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Original Paper

Designing Supportive e-Interventions for Partners of Men With Prostate Cancer Using Female Partners' Experiences: Qualitative Exploration Study

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Abstract

Background: Partners of men living with prostate cancer (PCa) can experience a variety of unmet needs that are largely unaddressed by health care professionals. There is limited evidence to suggest which approach may be most effective in supporting partners' unmet needs and further research is required to determine how to provide support to caregivers and how technology solutions can be designed.

Objective: This study aims to explore the experience of partners of men living with PCa and their perceptions of the potential role of information technology in supporting their needs.

Methods: A qualitative descriptive methodology using focus groups and phone interviews was used. Purposive sampling was used to recruit people attending a national conference supported by a national PCa organization. Interview guides were adapted from an existing evidence-based smartphone app for caregivers of people with colorectal cancer. Sessions were audio recorded and transcribed verbatim. A coding framework was developed, and transcripts were coded line by line into the framework. Codes within the framework were grouped into descriptive categories that were then developed into analytical themes.

Results: A total of 17 female partners participated in the study, with an average age of 64 (SD 8.5) years. The following two main themes emerged: In the first theme, that is, *How technology can be shaped to support female partners of prostate cancer survivors*, the content and design of the smartphone app was discussed in addressing female partners' needs. The following four subthemes were developed: getting support from social networks and resources, the lack of relevant information, demystifying future care expectations during and following a PCa diagnosis, and delivering the smartphone app—to whom and from whom. In the second theme, that is, *The benefits and barriers of technology*, the suitability of smartphone apps as a supportive modality for female partners was described. This included three subthemes: the smartphone app as an appropriate modality for supporting female partners, the future anticipated benefits of using the smartphone app, and concerns for storing and accessing information on the internet.

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Conclusions: A smartphone app may be a suitable modality for providing information and peer support to female partners of men living with PCa. There is a need to provide peer support for female partners in future interventions to ensure that female partners' intimacy and daily practical needs are met.

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KEYWORDS

prostate cancer; prostatic neoplasms; e-intervention; smartphone; qualitative research; caregivers; mHealth; mobile phone

Introduction

Background

Globally, prostate cancer (PCa) is the most commonly occurring cancer in men [1], with over 16,000 men diagnosed in Australia in 2020, accounting for approximately 20% of all male cancer diagnoses [2]. PCa survivorship research has predominantly focused on the psychological and physical effect of PCa treatments on men [3]. The psychosocial impact on their partners is an emerging area of priority, with research suggesting they may experience greater levels of distress than the individual with PCa [4-6], which may be attributed to avoidant communication between patients and partners [7]. Partners also report their own needs often go unaddressed by health care professionals [8]. As evidence of acceptable and effective interventions to support the partners of men with PCa remains unclear [9], further understanding of partner- or caregiver-specific issues is increasingly recognized as important to inform evidence-based supportive interventions [3,9]. A clearer understanding is needed to determine whether e-interventions can be adapted to meet partners' specific needs.

Dyads refer to the patient and spousal partner, and dyadic interventions remain an area of uncertainty within PCa. For instance, interventions are patient focused and produce conflicting results between patient and partner outcomes [9]. Unmet needs refer to areas where support may be lacking [10] and often can be organized several categories, including access to services, psychological care, financial support, relationships and communications, information, and spirituality [11]. Caregivers of people with cancer in general experience unmet needs related to providing symptoms and side effects management and maintaining function and caring for themselves [12]. Caregivers often experience elevated levels of distress across the disease trajectory [13]—less than 40% of caregivers participate in social events [14] and many experience financial burdens associated with loss of employment related to changing health status of the cancer survivor or increased caregiving responsibilities at home [15]. Previous studies have identified that there are over 200 unmet needs or issues that caregivers may experience [16]. Although there is the requirement to deliver interventions for the most distressing needs of caregivers, it is also imperative to provide caregivers with access to support and resources to address their less common needs [17]. Although flexibility in technology designs provide the potential to meet a range of unmet needs experienced by partners of men with PCa by having the capacity to tailor programs to users' needs, how best to design and deliver e-interventions requires more investigation.

e-Interventions and smartphone apps in particular have the potential to deliver resources to large groups of people [18] and facilitate the delivery of individually tailored care. The majority of Australians currently own smartphones and use of smartphones app is expected to increase [19]. Smartphone apps offer flexibility when seeking information and support as they allow caregivers to locate resources privately and from anywhere in the world with an internet connection [20]. e-Interventions have previously been used among caregivers of people with cancer and provide promising results; however, there is limited information about the use of smartphone apps [21]. A previous pilot study of a smartphone app for caregivers of people with colorectal cancer found that smartphone apps can be useful for caregivers when managing their own needs and that resources such as this should be available to all caregivers in a similar situation [22]. To be beneficial, smartphone apps should be highly relevant and appropriate to the needs of caregivers and easy to access [22,23].

A smartphone app has been developed using a user-centered design approach for caregivers of people with cancer [24] and trialed among caregivers of people with colorectal cancer [22]. This smartphone app was found to be feasible for caregivers and acceptable, with 85% of caregivers stating that they think the smartphone app should be made available to other caregivers looking after another adult with cancer [22]. This smartphone app, called Carer Guide, addressed unmet needs that caregivers commonly experience such as cancer information, including diagnosis, treatment, side effects, and symptoms; their mental well-being; lifestyle tips for caregivers, including diet and exercise; financial allowances and legal tips; and hospital contacts and information. Currently, no smartphone apps exist to support caregivers looking after men with PCa and the needs of these caregivers in relation to a technology solution is unclear. Therefore, this study aims to explore the experiences of partners of survivors of PCa as caregivers and their perceptions of the potential role of a smartphone app in supporting their unmet needs across the stages of PCa diagnosis.

Objectives

Using a qualitative study design, the objectives of this study are as follows:

1. Explore the experiences of partners of survivors of PCa as caregivers and how a smartphone app may support their unmet needs.
2. Identify partner preferences around the potential role of smartphone app in supporting their needs.
3. Obtain feedback on an example smartphone app and the potential role of a generic platform that can incorporate

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different aspects of PCa disease progression during the life course of survivors of PCa.

Methods

Setting

This study included two parts: (1) one focus group at an Australian national conference day held in Brisbane, Queensland, for PCa and (2) phone interviews with partners of men with PCa recruited nationally from an Australian PCa organization registry. The first part was conducted in July 2019, and recruitment for the second part occurred between May and June 2020. This study received approval from the Human Research Ethics Committee of the University of Technology Sydney (ETH19-3700) and Deakin University (2019-244).

Participants

Purposive sampling was used to recruit eligible partners.

Inclusion Criteria

Inclusion criteria included eligible partners (male or female) of men diagnosed with PCa at any stage of the disease and aged ≥18 years.

Exclusion Criteria

Exclusion criteria included people who were unable to follow conversations in English language.

In the first part, recruitment flyers advertising the date and location of the focus group were disseminated through the networks and caregiver support groups of the Australian PCa organization. In the second part, a recruitment flyer was released within the Australian PCa organization registry of people willing to be involved in research. Partners interested in participating in the project initiated contact with the research team (NW or AG) by phone or via email. Interested partners were emailed a copy of the plain language and consent form that was either signed on the day of the conference or returned via email before phone interviews.

Data Collection

This study was conducted using a qualitative descriptive design [25], informed by the epistemology of pragmatism that seeks to ascertain whether the knowledge generated has served the specific purpose of the study [26]. The focus group was facilitated by NW, AG, and PML and was attended by one nurse counselor as an observer to the group. The nurse counselor was an attendant at the conference and was external to the research project. The focus group was held in a quiet room away from the main conference. The phone interviews were conducted by NW. The focus group and phone interviews were guided by the same semistructured questions with prompts. The inclusion of phone interviews allowed us to invite people living across Australia and allowed the research to continue during COVID-19 pandemic lockdowns and when partners were unable to attend focus group sessions. The same questions and prompts were used in both focus groups and phone interviews to ensure consistency across sessions. The questions had been used in a previous study aimed at developing a smartphone app for caregivers of people with cancer [24,27].

Partners were asked questions about their experiences with PCa, how they found support, and the suitability of smartphone apps to meet their unmet needs. To support discussion, screenshots of an existing smartphone app for caregivers of people with colorectal cancer [24] (Figure 1) were shown to partners who were then asked to respond to questions about how the smartphone app may be specifically adapted to meet the needs of partners of men with PCa. The original smartphone app *Carer Guide* was developed using a user-centered design approach [28] and provided a source of information and resources specific to caregivers identified unmet needs. Primarily *Carer Guide* was a static source of information with supporting resources, including notepad and contacts for caregivers to enter their information as required. The potential to expand the smartphone app to include tailored features to engage with users is the aim of this study. The focus group and phone interviews were audio recorded and transcribed verbatim. Data collection continued until data saturation occurred.

Demographic data were collected from partners who participated in phone interviews. Partners who attended the focus group were invited to complete a demographic questionnaire; however, this was not mandatory. Completed data were collected for 11 partners.

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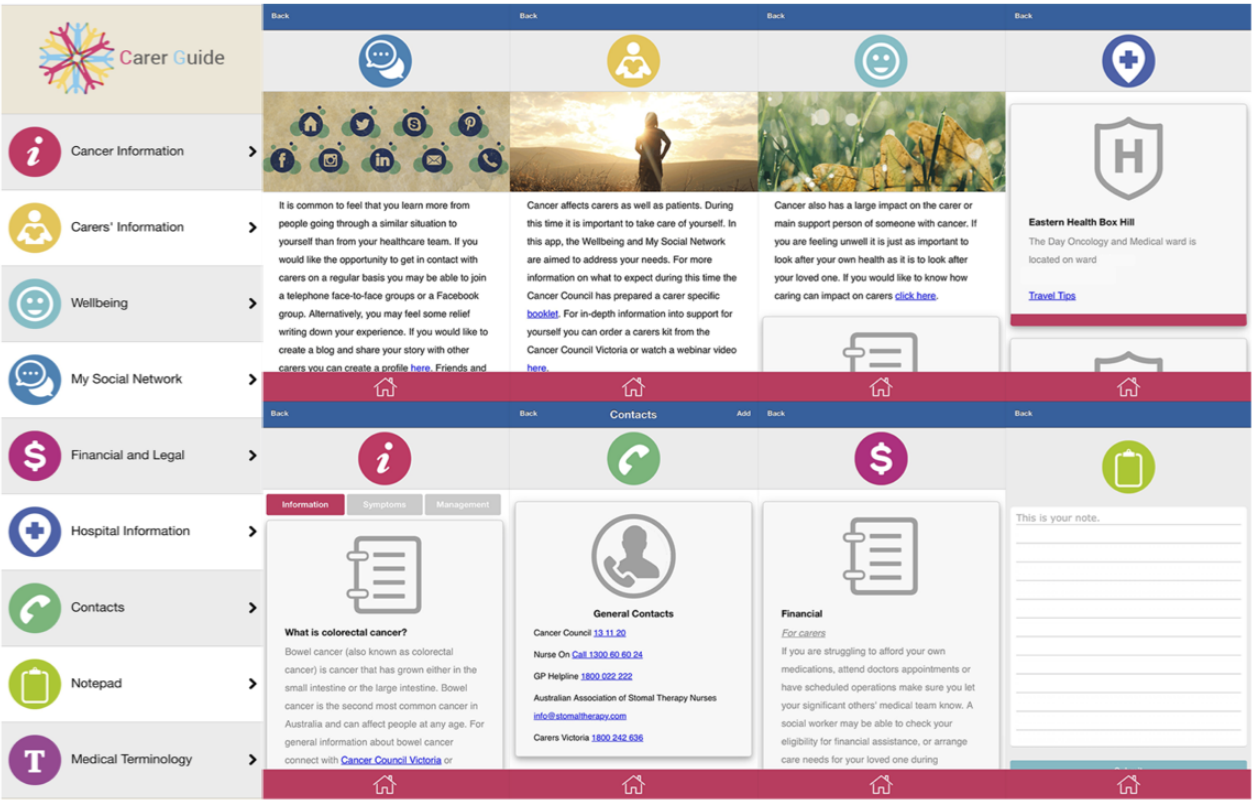
Figure 1. Previous smartphone app (Carer Guide) with content displayed for partners to review: Cancer Information, Carer Information, Wellbeing, My Social Network, Financial and Legal, Hospital Information, Contacts, Notepad, and Medical Terminology.



Data Analysis

Overview

Transcripts were read twice by 2 researchers (NW and AG) and a framework analysis was used based on initial impressions of overarching concepts [29]. Two authors (NW and AG) independently developed key codes; the authors then discussed the codes and agreed upon the codes that would be included in the framework. Transcripts were then coded line by line using NVivo software (QSR International) into the coding framework by one author (NW) and were checked by a second author (AG). Codes were grouped into similar and contrasting descriptive categories, which were then developed into themes and subthemes with interpretation confirmed by the full authorship team. Concepts were similar between focus group and phone interview transcripts, and thematic analyses were subsequently combined. Demographic data were analyzed using descriptive statistics. This study followed the COREQ (Consolidating Criteria for Reporting Qualitative Research) guidelines for qualitative studies [30].

Rigor

To ensure trustworthiness of the findings the following steps were taken as recommended by Bradshaw et al [31]. Credibility was promoted by establishing a trusting relationship and rapport with partners during the consenting process before conducting focus groups and phone interviews, and empathy was provided during sessions [31]. To provide confirmation of results, field notes were taken during focus groups and phone interviews to confirm major and minor themes from the thematic analysis process [31]. Demographic information was collected from partners where possible, and direct quotes were used to demonstrate findings [31]. An audit trail was used to provide dependable study procedures and results. To enhance the transferability purposive sampling was used [31].

Research Team

The research team consisted of psycho-oncology researchers with backgrounds in nursing, psychology, and social science who cumulatively had over 50 years of experience in providing support to people with cancer.

Results

Demographic Characteristics

A total of 17 female partners participated in the study—8 (47%) in the focus group and 9 (53%) in phone interviews. The average age of female partners was 64 (SD 8.5) years, and all were living with their male partners diagnosed with PCa. Complete demographic data are given in Table 1. Of the 9 partners in phone interviews, 8 (89%) provided information about the state they resided in Australia: 62% (5/8) lived in New South Wales, 12% (1/8) in Victoria, 12% (1/8) in Queensland, and 12% (1/8) in Western Australia. The focus group ran for 112 minutes, and phone interviews were on average for 47 (SD 12; range 31-67) minutes.

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Table 1. Demographic data of female partners (n=11).

Characteristics	Value
Age (years), mean (SD)	64 (8.5)
Female, n (%)	11 (100)
Residing with patient, n (%)	11 (100)
Highest level of education, n (%)	
Secondary school	2 (18)
Certificate or diploma	4 (36)
University degree	4 (36)
Postgraduate degree	1 (9)
Current or past caregiver, n (%)	
Current	8 (72)
Past	3 (27)
Length of time in the caregiver role, n (%)	
6 months to 1 year	1 (9)
1 year to 2 years	1 (9)
>2 years	9 (81)
Treatment received by the partner, n (%)	
Hormone therapy	5 (45)
Not currently receiving treatment	5 (45)
Drug trial	1 (9)
Self-identification as a caregiver, n (%)	
Yes	6 (54)
No	5 (45)

Findings

Overview

In addition to concepts already outlined in the *Carer Guide* app (Figure 1), female partners suggested content and resources that could support partners across the PCa survivorship continuum. Two overarching themes emerged from the data: *how technology can be shaped to support female partners of PCa survivors* and *the pros and cons of technology*. An overview of the themes and subthemes is given in Textbox 1.

Textbox 1. Themes and subthemes derived from the thematic analysis.

How technology can be shaped to support female partners of survivors of prostate cancer

- Support from social networks and resources
- The lack of relevant information
- Demystifying future care expectations during and following a prostate cancer diagnosis
- Delivering the smartphone app—to whom and from whom

The benefits of and barriers to technology

- A smartphone app as an appropriate modality for supporting female partners
- The future anticipated benefits from using the smartphone app
- Concerns for storing and accessing information on the internet

How Technology Can Be Shaped to Support Female Partners of PCa Survivors

In this theme, female partners discussed how smartphone apps could be designed to meet their needs. Four subthemes emerged highlighting the unmet needs of female partners and included support from social networks and resources; the lack of relevant information; demystifying future care expectations during and following a PCa diagnosis; and delivery the smartphone app—to whom and from whom.

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Support From Social Networks and Resources

Informal social networks of friends and family members, as well as more formal networks of partner support groups, were the main source of support for female partners of men with PCa. Benefits from attending support groups included shared learning, emotional support for intimate and communication issues within the dyad as a result of the PCa diagnosis, and support for female partners' own individual needs:

> *They [women] tell you exactly what's happening to their husbands and their impotence and their sexual life. And women are very honest.* [ID3]

The inclusion of support networks within a smartphone app was, therefore, a key feature to include, for female partners in particular, to streamline and improve access to this source of support, which was, at times, difficult to find independently:

> *Initially finding information about support groups and things like that, that was quite hard to find.* [ID1]

In recognition of diverse preferences for sharing information among female partners, one woman also suggested the inclusion of videos of couples discussing their experiences of PCa may be useful for partners who preferred not to access information and support from support groups:

> *Hearing those two [information video of a couple affected by prostate cancer] talk about...their sex lives and how that had changed for them...I actually found that really useful actually hearing people talk about their experience...And you know, if you, if you didn't want to go off to a support group, or if you didn't have a support group available to you, just being able to sit down and even replay those interviews or absorb them at your own pace, I think was really, was really worthwhile.* [ID9]

Similarly, a gap in knowledge of available resources was evident. Female partners reported their ack of knowledge on what they entitled to, including access to Prostate Cancer Nurses and community resources, and identified that this could be addressed in an app by highlighting available services to female partners:

> *Where do you find out about how you can get some funded counseling? Where do you go?* [ID4]

The Lack of Relevant Information

Female partners described information related to the diagnosis of PCa, treatment, and treatment outcomes was lacking. Often, side effects of treatment were understated and the lasting side effects of hormonal therapy were poorly explained by clinicians and information sources:

> *There might be sexual dysfunction...You know it's like just a little dot point on the page when...I feel like that should really be highlighted more for partners.* [ID9]

Some female partners expressed frustration at the representation of PCa as a *safe cancer*, as this was not their experience, and the lack of available information was insufficient to meet their understanding:

> *I would have liked to have understood better just how big a risk he was at...I don't think that that's made very clear...You know Gleason 9 is like exponentially worse than Gleason 7...I don't think I appreciated how much at risk he was for recurrence.* [ID4]

Female partners also noted that information should be specific for couples at different stages in their life, such as those still wanting to have children later in life:

> *Some partners may, particularly if they're younger...there would have to be a whole message about you need to go and...get sperm frozen.* [ID9]

Having had access to a smartphone app to find PCa-specific information was noted as a valuable resource that could have saved time, improved access to information, and reduced anxieties of female partners:

> *I think it [having an app] would just expedite the information gathering. Having a one stop shop.* [ID3]
>
> *I would still have had the anxiety about how successful his treatment is going to be. But if I had a bit more understanding of what was going...[it] would have taken a bit of the edge off in terms of the anxiety.* [ID2]

Demystifying Future Care Expectations During and Following a PCa Diagnosis

Female partners identified the need for clear information on what to expect and what care they would need to provide following their partners' diagnosis. This often included practical tips that were learned through informal networks or from experience:

> *I found a couple of people that I've spoken to, is where to get supplies from, like incontinence pads and how they cope with catheters when they get home.* [FG]
>
> *We just sort of did a lot by ear...I looked at my husband when he was in hospital and thought you won't be able coming out in jeans. I need to go and get tracksuit pants and ran off to the shops and bought tracksuit pants.* [ID1]

Female partners also felt that the app may provide them with information about what partners can expect at each stage of the cancer journey:

> *He'll have his own journey, he'll be doing his own thing, but this [information provided] is what you...can expect over the next you know, 3 months, 6 months, 12 months, 2 years, 3 years.* [ID9]
>
> *[Including] a what to expect kind of tabs...'cause at the outset that's really important.* [ID2]

In particular, some female partners noted wanting to feel prepared for end-of-life care and what would happen when the health of their partner deteriorated or how to manage future funeral arrangements:

> *Can I cremate him? You know, it took me three weeks to find an answer. No, you can't. You can't cremate him straight away. You've got to wait; I think it was*

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> *20 days or something like that...because of the radiation [from brachytherapy].* [FG]

Delivering the Smartphone App: To Whom and From Whom

There was variability around who female partners felt the main audience of the smartphone app should be. Some female partners felt that the smartphone app should be tailored specifically to partners:

> *I think it might be good to have an app just for carers actually, because then you're focusing on them...not the person with cancer and sometimes it is the carers who, who needs the help.* [ID7]

Alternatively, some female partners reported that the smartphone app should be delivered to both partners and patients or that both people in the relationship should have access to all of the information:

> *Maybe it will be better to have it for carers and patients together. Because then I'm a nosey person and I want to know what's going on.* [ID1]

The name of the smartphone app had an impact on female partners' perception of whether the smartphone app was meant for them. Some female partners responded positively to the term *caregiver*, whereas others felt that this did not match their experience:

> *I would think oh that's probably going to apply to people you know who are much older than me. That's what I think of when I hear the word carer.* [ID9]

Female partners agreed that the smartphone app should be supported or recommended by reputable sources to encourage them to download it, such as health care professionals, cancer organizations, or advertisements within hospitals:

> *If it was sitting there in the waiting room [outpatient oncology], I would have picked it up and I, I would have looked at it. I would have logged on and checked it out.* [ID2]

Benefits of and Barriers to Technology

In this theme, the suitability of apps as a modality to support the female partners of men with PCa was explored. The three subthemes that emerged around are the smartphone app as an appropriate modality for supporting female partners, the future anticipated benefits from using the app, and concerns around storing and accessing information via internet.

A Smartphone App as an Appropriate Modality for Supporting Female Partners

Female partners responded positively to the layout of the *Carer Guide smartphone* app because the app was easy to navigate and its design was suitable for people looking after someone with cancer:

> *It's colorful, it's bright, it's cheery.* [ID3]

> *I like the menu, it looks really easy to navigate.* [ID9]

The purpose of the app was clear, and the original content covered aspects of care that female partners felt were important in supporting caregivers of men living with PCa:

> *I think that [a smartphone app] would be a help for a lot of people especially when their partners are newly diagnosed it's very difficult to understand it all. If there was one [smartphone app] just for carers that would give them some information that's specifically to them...it would have been helpful...It [the Carer Guide] goes through quite a few things that I think are interesting yeah, I think that you need to know.* [ID7]

Many female partners stated that they kept medical records and contact details separately, either in paper files or within regular mobile phone functions. General consensus among female partners was that storing this information within the smartphone app would be beneficial for quickly and easily locating information during emergencies, creating digital notes rather than numerous paper documents, and compiling all relevant information at the same location:

> *I think it's great that you can have contact numbers in there. Um, I struggle sometimes with finding, you know, who do you ring...especially if it's at 11 o'clock at night, and there's lots of pain.* [FG]

> *I can see how this [the app] could be useful and give you some way to put everything that it wasn't um it wasn't just all over the place...one of the things that everybody tells you as soon as you get diagnosed is start a binder with all your results...in some ways this [the app] could potentially complement that as a digital binder.* [ID4]

The use of reminders was predominantly discussed for daily care tasks such as medication and appointment reminders. One woman described that reminders and bookmark functions could also be used for her own needs when allocating time to visit certain sections of the app:

> *You could kind of do a follow up with the app...set yourself a reminder that something pops up somewhere in your calendar to say hey, don't forget to read this section you know you bookmarked this for yourself.* [ID9]

Another female partner expressed that storing this information within the *Carer Guide* smartphone app would be preferable and beneficial for her mental well-being compared with other smartphone apps available on her phone:

> *We put all that stuff [scheduling] in our outlook calendar, so then it pings at you and reminds you when you don't really want to know about it...That would be really nice to have in its own little world when you're in the right mind frame to go and look at it...Even as someone who's not actively dealing with those sort of treatment schedules...when we [are waiting to] get the results back...that's when I'm most on edge...so putting that appointment reminder somewhere else [in the Carer Guide app] would be nice.* [ID4]

Two female partners identified that they would be unlikely to use a smartphone app as a modality for support. Other female partners suggested that although they use smartphone apps,

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levels of technology literacy and comfort in using smartphone apps may vary for other female partners:

> *Well see I probably wouldn't use it very much.* [FG]
>
> *But I've never actually gone into the app store and just put in the word prostate to see if there's a – there may be something there. I – I don't know. That's not how I search for stuff.* [P1]
>
> *Well, it's because we're not used to doing that, these old people...these apps are very new.* [P2]

The Future Anticipated Benefits From Using the Smartphone App

Reflecting on how access to the *Carer Guide* smartphone app may have supported them in earlier stages of their caregiver journeys, female partners identified a number of potential benefits. Benefits included having the app as a *one-stop shop* for everything that they needed while *on the run*. In the context of providing care to their partners with PCa, female partners identified that the smartphone app would have been a useful source of information to understand more about what was happening and what they needed to learn more about:

> *It would have given me a good structure to start knowing what I needed to get organized and potentially what I needed to start asking questions on.* [ID4]

Female partners also felt that access to the smartphone app would have provided them with the opportunity to seek information to address their own needs without the patient knowing about it:

> *[in reference to whether the app would help in finding information for caregivers] Definitely, definitely. 'cause sometimes I would actually feel a bit guilty for looking up stuff for myself.* [ID2]

Concerns for Storing and Accessing Information via the Internet

Some areas of concern when using technology included the security of personal information stored on the internet and hesitation toward possibilities of accessing overwhelming information that can lead to distress. The ability to input medical information or link the smartphone app with medical record platforms such as MyGov was noted as benefits of the smartphone app for storing up-to-date medical information. However, concerns over risk of web-based information breach was noted among female partners:

> *What about keeping a medical record?...The app would have to be very secure.* [FG]

Similarly, female partners noted that the inability to understand overwhelming medical content available on the internet along with lack of clinical support to explain the web-based content was a potential concern for some partners:

> *When you're given a booklet by the doctor...it's actually an opportunity for somebody to discuss it. Whereas when you go searching for stuff on the Internet...if you find too much in-depth information it can panic you.* [ID5]

Discussion

Principal Findings

Overall, female partners of men living with PCa responded positively to the potential use of a smartphone app as a modality of support during their cancer journey. A previous smartphone app designed for caregivers of people with colorectal cancer, *Carer Guide* [21], was acceptable to female partners, and the original content (cancer information, carer information, well-being, social network, hospital information, financial and legal, and medical terminology) was appropriate to their needs. Content specific to the shared experiences of these female partners and additional content that may be helpful to include in future e-intervention designs included more prominent linking with peers and support groups, information and support related to the effects of PCa treatment on their intimate relationships, and more clarity on what to expect during each stage of the caring period. The need for flexibility with future interventions was apparent, as some female partners preferred to have access to a smartphone app specifically for caregivers, whereas others thought it should be a resource shared between the patient–caregiver dyad. There is a need to ensure users are aware of the level of security that can be offered by digital health devices, particularly when they are being used to input or store medical information to ensure privacy and confidentiality.

Addressing the patient–partner dyad intimate relationship during a PCa diagnosis is a primary area of focus within the literature [32]. Female partners in this study again highlighted the impact of PCa on their intimate relationship and their ability to seek support for themselves, suggesting this is as an ongoing unmet need. Dyadic research has shown that partners' needs as caregivers in the literature are not well-addressed in the PCa field [9]. Previous studies have described that strong couples' communication can have positive effects on mental outcomes of patients and caregivers [33]. However, in this study, female partners also highlighted the need for open communication with others such as wanting practical advice from peers about how to manage frustrations with intimacy on a daily basis.

Female partners reported that practical support was also required for daily care tasks and needed incorporation into the smartphone app. There is evidence that caregivers are performing clinical tasks in the home setting [34]. However, the need for guidance on daily tasks, such as locating incontinence products, preparing for surgical recovery, and managing day-to-day needs is apparent, and there is little in the literature to suggest that these types of unmet needs are being addressed or that there are effective symptom management interventions. The need for peer support was a key approach female partners felt intimate and practical issues could be addressed. Peer support can help caregivers to find information and resources related to patient care and share emotional experience with others in a similar situation [35]. These female partners demonstrated this through the sense of belongingness and empathy when talking about intimate issues with each other. Providing a range of communication and supportive resources in the smartphone app may be more beneficial in meeting

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caregivers' unmet needs related to intimate and practical issues; however, this requires further investigation.

Female partners were favorable to the concept of using a smartphone app while caring for a male partner with PCa. However, there are varied perspectives on whether resources such as Carer Guide are feasible for use for all caregivers because of personal preference for seeking information. From our previous work, it was evident that caregivers required the content and functionality of a digital resource to be specific to their situation to fulfill their unmet needs [27]. In this study, female partners requested specific information related to different stages of disease and when disease progression occurs during a PCa diagnosis. Female partners noted having information would reduce fears of the unknown and potentially reduce associated anxiety. Of note, some female partners particularly mentioned seeking information about bereavement care or care after death, suggesting that female partners would like information to feel prepared for this period of care. Unmet needs related to palliative care have previously been recorded in a systematic review and are present in 21% to 100% of participants across studies, suggesting that there is great variability in the need for information about palliative and bereavement care [36]. However, only one of these studies was within the PCa caregiver field [37], suggesting that more research is needed in this area.

An additional finding of this study was the need for flexibility in the audience of an e-intervention. PCa in particular is often seen as a couples' disease [38]; therefore, there is a requirement to provide support for the couple going through a PCa diagnosis. However, as described, female partners can experience both unmet needs specific to their caring role and needs different from the dyadic relationship. Therefore, these individual unmet needs also require tailored support. A recent systematic review by Luo et al [39] recommended that interventions for families affected by cancer should include four components: information, communication and support, skill building, and psychoeducation. It is important, therefore, to encompass all of these domains when designing and developing interventions to enhance their potential to improve caregivers' outcomes. Flexible designs or the ability to customize items are required to ensure apps can be tailored to caregivers' unmet needs. Furthermore, as identified by Lambert and Girgis [17], it is necessary for e-interventions to address less common unmet needs of caregivers, highlighting the importance of flexibility in design, content, and delivery of interventions for caregivers.

In creating reliable e-interventions for caregivers, it is also paramount to consider the safety and privacy concerns of end users. As described by the female partners in this study, when entering medical or sensitive information into smartphone apps, it is vital that users are confident that they know how their information is being stored and with whom it is being shared, if anyone. Several frameworks exist to assess the different functionalities of smartphone apps. Variance in these frameworks can lead to difficulties evaluating smartphone apps [40]. To ensure the safety and privacy of end users across trials, standardized approaches for the assessment and production of smartphone apps are required. With more streamlined e-interventions, it may be possible to provide caregivers with the opportunity to engage with a greater number of web-based resources to meet their needs.

Within the clinical setting, it is of vital importance that e-interventions are supported by health care professionals during implementation into routine practice. Referrals and recommendations to existing and up-and-coming interventions are required by clinicians in outpatient and general practice settings to support partners and caregivers who regularly prioritize their own unmet needs last [41].

Limitations

This study had several limitations, including the small sample size and homogeneity in participation of only female partners. The study was advertised for both male and female partners to participate; however, no male partners initiated contact. Furthermore, description of the study sample was limited, as not all partners who participated completed demographic questionnaires. Despite the small sample size, findings were similar to previous research describing partners' experiences of supporting men with PCa [8]. Partners in our sample lived in several states across Australia, suggesting that experiences were similar across the country; however, this should be interpreted with caution because of the small sample size, as these findings may not be transferable to other demographic context. Future research should endeavor to assess experiences, and the adaptation of support modalities in same sex attracted men affected by PCa.

Partners were shown screenshots of the smartphone app rather than having the opportunity to use the app prototype. This may have resulted in different consequences about the usefulness of the app. However, the main intent of this study was to understand how the content could be extended to the PCa setting. In addition, 2 authors (NW and PML) were the original developers of the Carer Guide smartphone app. To reduce any potential biases in findings, multiple members of the authorship team, including those with expertise in PCa survivorship and those with no prior involvement in the original smartphone app, were involved in the adaption of the interview guide and each stage of data analysis.

Conclusions

A smartphone app may be a suitable modality for providing information and peer support to female partners of men living with PCa. A few changes are required to adapt an existing smartphone app to partners' specific needs. There is a strong need to provide peer support for female partners in future interventions to ensure that female partners' intimacy and daily practical needs are met.

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Conflicts of Interest

None declared.

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Abbreviations

COREQ: Consolidating Criteria for Reporting Qualitative Research
PCa: prostate cancer

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Review

The Extent of Engagement With Telehealth Approaches by Patients With Advanced Cancer: Systematic Review

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Abstract

Background: Telehealth approaches are increasingly being used to support patients with advanced diseases, including cancer. Evidence suggests that telehealth is acceptable to most patients; however, the extent of and factors influencing patient engagement remain unclear.

Objective: The aim of this review is to characterize the extent of engagement with telehealth interventions in patients with advanced, incurable cancer reported in the international literature.

Methods: This systematic review was registered with PROSPERO (International Prospective Register of Systematic Reviews) and is reported in line with PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) 2020 guidelines. A comprehensive search of databases was undertaken for telehealth interventions (communication between a patient with advanced cancer and their health professional via telehealth technologies), including MEDLINE, Embase, CINAHL, PsycINFO, Cochrane Library, Sociological Abstracts, and Web of Science, from the inception of each electronic database up until December 31, 2020. A narrative synthesis was conducted to outline the design, population, and context of the studies. A conceptual framework of digital engagement comprising quantitative behavioral measures (frequency, amount, duration, and depth of use) framed the analysis of engagement with telehealth approaches. Frequency data were transformed to a percentage (actual patient engagement as a proportion of intended engagement), and the interventions were characterized by intensity (high, medium, and low intended engagement) and mode of delivery for standardized comparisons across studies.

Results: Of the 19,676 identified papers, 40 (0.2%) papers covering 39 different studies were eligible for inclusion, dominated by US studies (22/39, 56%), with most being research studies (26/39, 67%). The most commonly reported measure of engagement was frequency (36/39, 92%), with substantial heterogeneity in the way in which it was measured. A standardized percentage of actual patient engagement was derived from 17 studies (17/39, 44%; n=1255), ranging from 51% to 100% with a weighted average of 75.4% (SD 15.8%). A directly proportional relationship was found between intervention intensity and actual patient engagement. Higher engagement occurred when a tablet, computer, or smartphone app was the mode of delivery.

Conclusions: Understanding engagement for people with advanced cancer can guide the development of telehealth approaches from their design to monitoring as part of routine care. With increasing telehealth use, the development of meaningful and context-

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and condition-appropriate measures of telehealth engagement is needed to address the current heterogeneity in reporting while improving the understanding of optimal implementation of telehealth for oncology and palliative care.

Trial Registration: PROSPERO (International Prospective Register of Systematic Reviews) CRD42018117232; https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42018117232

(JMIR Cancer 2022;8(1):e33355) doi:10.2196/33355

KEYWORDS

systematic review; advanced cancer; engagement; digital health; telehealth; mobile phone

Introduction

Background

Cancer ranks as a leading cause of death worldwide and is a leading cause of premature death in most countries [1]. For people living with advanced cancer, fluctuating unmet needs can be experienced over time with disease progression [2]. Common symptoms include pain, experienced in approximately two-thirds (66.4%) of patients with advanced disease [3], alongside breathlessness, nausea and vomiting, and fatigue [4]. Typically, individuals experience more than one symptom, with an average of 14 symptoms for those with advanced cancer [5]. Such physical symptoms often exist alongside deterioration across physical, psychological, social, spiritual, and overall quality of life (QOL) trajectories [6]. There remain gaps in supporting care delivery for patients with cancer, including barriers in health communication with health care providers, lack of care coordination, and challenges in accessing care [7].

Telehealth and telehealth interventions refer to a method in which the patient and health care professional can communicate clinical information remotely via a number of different mediums such as telephone, web-based methods, and mobile apps [8]. This method is increasingly used to deliver cancer care as it provides opportunities for efficient and flexible service delivery and enables clinicians to maintain involvement independent of the physical location of the patients or clinicians [9-12]. These characteristics have also driven their increased application to support delivery of care during the COVID-19 pandemic, enabling avoidance of direct physical contact while contributing to provision of continuous care in the community. Telephone-based approaches have been highlighted as a possible means of overcoming gaps in service delivery for patients with cancer [7], including reducing the travel required to access support services that can lead to physical, psychological, and financial stress [13,14]. Examination of telehealth approaches for patients with chronic diseases has found varying effects, with improved self-management of diabetes and reduced mortality and hospital admissions in heart failure, but these improvements have not been observed across other conditions, including cancer [8]. Emerging evidence is mixed, with a recent review that focused on all cancer stages demonstrating clinical equipoise, with no discernible difference between telehealth and usual care in improving QOL [15]. However, a recent systematic review focusing specifically on patients with advanced cancer and diverse web and technological interventions (largely providing psychosocial, self-management, and expert-guided support) found that most approaches suggested some degree of efficacy relating to QOL and psychosocial well-being [16]. However, we do not know how well people with advanced cancer engage with these interventions.

With emerging clinical validation demonstrating the potential of digital technology approaches to improve care and outcomes of patients with advanced cancer, usability must also be considered [17]. Subjective aspects of usability require a better understanding, specifically regarding user satisfaction and engagement [17]. Patient engagement can be an important factor in the success of health interventions, leading to better intended health outcomes for the patient and lower health care costs [18]. As such, the effectiveness of telehealth interventions in improving health outcomes is heavily dependent on patient engagement. However, patient engagement is a broad term that can cover multiple levels of how a patient interacts with an intervention. For the purposes of this review, with a focus on technology-based interventions, *engagement* will be used to refer to the specific quantitative measures of behavior of engagement as defined by Perski et al [19] (ie, comprising the frequency, amount, duration, depth of use, and other measures of use and interaction with a digital health intervention). A previous systematic review found that information technology platforms (eg, mobile phone devices, internet-based interventions, social media, and other web-based communication tools) can help engage patients in health care processes and motivate health behavior change [20]. However, interventions with the intention to help support patients in managing chronic conditions can be complex. There is a need to understand whether different aspects of telehealth interventions uniquely influence patient engagement, especially for patients with advanced cancer who often experience a high symptom burden and functional impairment [21]. Understanding patients' engagement with telehealth interventions is necessary to further evaluate and refine the implementation of these emerging and promising approaches for patients with advanced cancer. Therefore, there is a need to understand how patients with advanced cancer engage with telehealth interventions and which aspects of these interventions may influence engagement.

Objectives

Past systematic reviews have sought to synthesize the evidence of telehealth interventions among patients with cancer and survivors but have not explored interventions solely intended for and tested on patients with advanced, incurable cancer [15,16]. Understanding patient engagement can help us evaluate and refine further design, development, and evaluation of telehealth approaches for people with advanced cancer. A companion review [22] explored the clinical and cost-effectiveness of the interventions on health and health

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system outcomes, whereas this review synthesizes the data on patient engagement with the interventions. The aims of this review are as follows: (1) to characterize the extent of behavioral engagement of people with advanced, incurable cancer with telehealth interventions and (2) to explore factors that influence engagement with telehealth interventions.

Methods

Information Sources

This systematic review was registered with PROSPERO (International Prospective Register of Systematic Reviews; CRD42018117232). A systematic review of the literature was conducted in the following databases: MEDLINE, Embase, CINAHL, PsycINFO, Cochrane Library, Web of Science, and Sociological Abstracts, with studies included from the inception of each electronic database up until December 31, 2020. No lower cutoff date was chosen as there has not been a previous review looking into engagement with telehealth interventions in this population. An example search strategy used for MEDLINE can be found in Multimedia Appendix 1 and includes keywords and medical subject headings. The development of the search strategy was supported by information specialists at the University of Leeds. This search was supplemented by forward and backward citation searching of key papers. This review was reported in line with the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) 2020 guidelines. The Centre for Reviews and Dissemination guidelines directed our process for conducting this systematic review and the decisions made [23].

Eligibility Criteria

Studies were eligible for inclusion in the review if the following applied:

1. They involved a telehealth intervention, which is defined as "any intervention in which clinical information is transferred remotely between patient and health care provider, regardless of the technology used to record or transmit the information" [8]. This could include symptom measuring or monitoring (eg, Patient-Reported Outcome Measures); education, information giving, and support, including decision aids and advanced care planning; psychological interventions; or medical consultation (telemedicine or teleconsultation). Participants could be located anywhere as long as the intervention that was carried out conformed to the telehealth definition.
2. They included participants of any age who were living with cancer of any type that could not be cured (advanced, metastatic, or terminal). This included people who had been treated with curative intent but whose cancer had recurred or progressed, those not being treated with curative intent, and those at or near end of life.
3. They included a measure of engagement as an outcome or reported as part of the study findings. In this review, we used the measures conceptualized as behavior that were identified by Perski et al [19]: frequency, amount, duration, and depth of use.
4. The studies were carried out in any country at any time.
5. Risk of bias was not used as a selection criterion for inclusion in the review.

Studies were excluded when the following applied:

1. The participants included patients with cancer currently being treated with curative intent, and the studies had mixed populations (ie, not 100% of the sample were people with cancer that could not be cured), unless findings pertaining to our population of interest were presented separately in the results section.
2. The studies did not report primary data (eg, systematic reviews, study protocols, conference abstracts, editorials, and commentaries).
3. The studies were not in the English language.

Study Selection and Data Collection Process

In total, 2 authors (WG and MA) reviewed titles, abstracts, and full-text papers, assessing them for eligibility independently. Any disagreements were resolved through discussion.

Data from the included studies were extracted into a predesigned form by WG and verified by MA to capture study characteristics (design, sample size, cancer type, gender, age, and outcomes). Data were also extracted based upon the items included in the Template for Intervention Description and Replication checklist (why, what, who provided, how, where, when and how much, tailoring, modifications, and how well) [24].

Quality Assessment

The included studies were assessed for methodological quality and risk of bias independently by 2 authors (WG and MA), with any disagreements resolved through discussion. The risk of bias for randomized controlled trials (RCTs) and nonrandomized studies was assessed using the Mixed Methods Appraisal Tool [25].

Data Synthesis

A narrative synthesis [26] was conducted to outline the design, population, and context (mode of delivery, health care provider, and intervention intensity) of the individual studies. Studies were categorized by their approach to examining intervention effect, differentiating between those exploring pure intervention effect (eg, using blinded RCT designs) and those exploring effect in the context of routine health care [27]. For the primary outcome of engagement, a deductive and inductive approach was taken using the definitions of engagement behavior outlined by Perski et al [19] while also ensuring that other engagement-related data were captured. Engagement data were identified and split into categories based upon the type of engagement the studies measured: frequency (how often contact was made with the intervention over a specified period), the amount or breadth (the total length of each intervention contact), duration (the period over which participants were exposed to an intervention), and depth (variety of content used) [19]. Across these 4 measures, studies were grouped together based upon how they measured the outcome, which was then summarized.

Data from the included studies relating to frequency of use by patients, where reported, were transformed to a percentage of actual patient engagement compared with intended engagement

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with the intervention to provide a standardized statistical comparison. When overall engagement percentages were calculated, these were weighted by sample size.

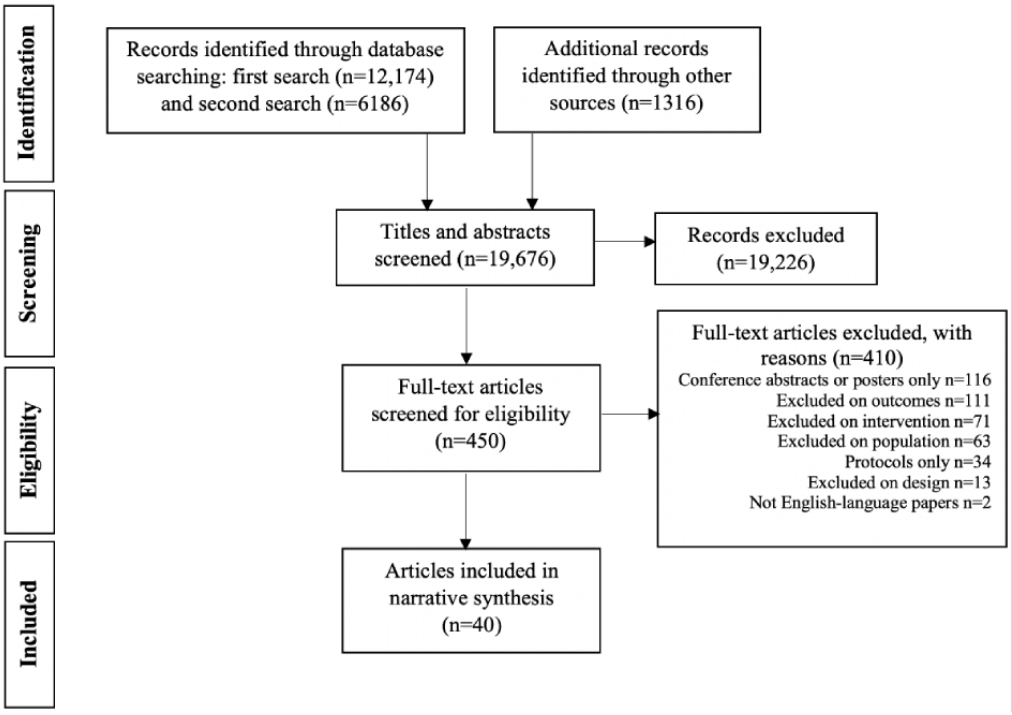
To draw associations between the calculated percentage of actual patient engagement, the intensity of the intervention (for the patient and health professional), and mode of delivery, we had to simplify these characteristics. The intensity of the interventions for both the patient and the health professional was coded by a member of the research team (WG). WG reviewed the intervention description in each included study to determine the expected engagement with the intervention for patients and health professionals. This referred to any interaction (both scheduled and unscheduled) that was anticipated or planned with the intervention (eg, a patient having a telephone consultation with a health professional or submitting data via a web-based system). For articles where a second opinion was requested by WG, a second reviewer (MA) discussed the study with WG until a consensus was achieved on the expected engagement reported. The expected engagement was simplified into categories of high, medium, and low expected engagement to make comparisons across studies. For patients, low expected engagement referred to only having ≤3 contacts with the intervention, a medium level of engagement was 4 to 7 expected contacts, and a high level of engagement was ≥8 expected contacts or more than daily reporting of symptoms. A previous study of engagement with a web-based mindfulness intervention identified similar levels of high and low participant engagement (low: 0-4 and high: 5-7); however, a third category was added for this review to account for the studies with >7 contacts [28]. For health professionals, the categories mirrored those for patients if the health professional was required to make contact with the patient (eg, low was ≤3 contacts, medium was 4 to 7 contacts, and high was ≥8 contacts). If the health professional was required to only make contact with the patient when prompted to do so by a patient's entry on a system or survey, it was coded as low contact on the part of the health professional. For each intervention, we also coded the mode of delivery (eg, telephone, smartphone, or web-based), including interventions where multiple modes were used. We were then able to look at associations between the mode of delivery, expected level of engagement (high, medium, or low for the patient and health professional), and the percentage of actual patient engagement with the intervention.

Results

Search Results

Of the 19,676 papers that were identified in the database search, 0.2% (40/19,676) of papers covering 39 different studies were eligible for inclusion in the systematic review [29-68]. Figure 1 outlines the PRISMA flow diagram for the included studies and the reasons for exclusion of studies.

Figure 1. PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow diagram.



Study Characteristics

Table 1 includes a summary of the characteristics of the included studies. Table 2 outlines the characteristics of the included interventions and the engagement outcomes. The included studies had a sample size ranging from 6 [61] to 766 [31] and included multiple RCTs (16/39, 41%) [30,31,33-35,37-39,43-45,48,50,57,63,67,68], with most studies being conducted in the United States (22/39, 56%) [29-31,33,34,37-41,43,45,46,48,50,54,57,59,60,66-68]. Of the 39 studies included in the review, 13 (33%) explored intervention effects in the context of routine care implementation [29,32,36,41,42,49,51,52,58,60-62,64], with the remainder exploring intervention effects often using a blinded controlled trial design.

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Table 1. Characteristics of the included studies (N=39).

Study	Country	Study design	Sample size	Type of cancer	Age (years)	Female participants, n (%)
Alter et al [29]	United States	Pilot	8	Colorectal	Range 59-79	5 (63)
Badr et al [30]	United States	RCT[a]	39	Lung	Mean 68 (SD 10)	29 (74)
Basch et al [31]	United States	RCT	IG[b]: 441; CG[c]: 325	Breast, genitourinary, gynecologic, or lung	IG: median 61; CG: median 62	IG: 257 (58); CG: 187 (58)
Bensink et al [32]	Australia	Feasibility	11	Advanced cancer, type NR[d]	Range 3-18	NR
Bouchard et al [33]	United States	RCT	192	Prostate	Mean 69 (SD 9)	0 (0)
Bruera et al [34]	United States	RCT	190	Advanced cancer, type NR	Median 58 (range 25-84)	128 (67)
Chambers et al [35]	Australia	RCT	189	Prostate	Mean 70 (SD 9)	0 (0)
Chavarri-Guerra et al [36]	Mexico	Observational study	45	Advanced cancer, type NR	Median 68 (range 33-90)	26 (58)
Cheung et al [37]	United States	RCT	39	Breast	NR	39 (100)
Cheville et al [38,39]	United States	RCT	516	Multiple myeloma, myelodysplastic syndrome, or lymphoma	Mean 66 (SD 11)	257 (50)
Chow et al [40]	United States	Feasibility	190	Advanced cancer, type NR	Median 68 (range 39-89)	94 (49)
Cluver et al [41]	United States	Feasibility	10	Advanced cancer, type NR	Mean 50 (range 26-61)	7 (70)
Dixon et al [42]	Canada	Feasibility	69	Advanced cancer, type NR	Mean 69	19 (28)
Donovan et al [43]	United States	RCT	65	Ovarian	Mean 57 (SD 9)	65 (100)
Eldeib et al [44]	Egypt	RCT	IG: 44; CG: 38	Colorectal or gastric adenocarcinoma	IG: mean 50 (SD 11); CG: mean 45 (SD 13)	IG: 28 (64); CG: 24 (63)
Flannery et al [45]	United States	RCT	IG: 30; CG: 15	Lung	IG: mean 66 (SD 8); CG: mean 61 (SD 9)	IG: 7 (41); CG: 5 (45)
Fleisher et al [46]	United States	Feasibility	22	Advanced cancer, type NR	Range 37-77	11 (50)
Fox et al [48]	United States	RCT	192	Prostate	IG: mean 71 (SD 8); CG: mean 71 (SD 9)	0 (0)
Fox et al [47]	Australia	Feasibility	15	Melanoma	26-49 years: n=4 (27%), 50-64 years: n=6 (40%), ≥65 years: n=5 (33%)	7 (47)
Friis et al [49]	Denmark	Feasibility	20	Lung	Median 70.5 (range 54-86)	7 (35)
Gustafson et al [50]	United States	RCT	IG: 144; CG: 141	Lung	IG: mean 62 (SD 11); CG: mean 61 (SD 10)	IG: 62 (50); CG: 59 (48)
Haddad et al [51]	Canada	Feasibility	IG: 102; CG: 118	Lung and others	IG: mean 62 (range 35-83); CG: mean 60 (range 31-87)	IG: 28 (50); CG: 25 (45)
Hennemann-Krauss et al [52]	Brazil	Observational study	12	Advanced cancer, type NR	Mean 68 (SD 9)	5 (42)
Keikes et al [53]	Netherlands	Feasibility	155	Colorectal	NR	NR

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Study	Country	Study design	Sample size	Type of cancer	Age (years)	Female participants, n (%)
Liu et al [54]	United States	Pilot	16	Ovarian	Median 58 (range 36-80)	NR
Nemecek et al [55]	Austria	Feasibility	15	Non–small cell lung cancer, melanoma, and pancreatic	Mean 50	NR
Rasschaert [56]	Belgium	Feasibility	11	Colorectal, gastric or esophageal, pancreatic, and cholangiocarcinoma	Median 57 (range 44-74)	6 (55)
Rose et al [57]	United States	RCT	210	Advanced cancer, type NR	40-60 (n=109); 61-80 (n=101)	69 (33)
Sardell et al [58]	United Kingdom	Feasibility	45	Glioma	Median 50 (range 23-69)	15 (33)
Schmitz et al [59]	United States	Pilot	7	Breast	Mean 61	7 (100)
Sherry et al [60]	United States	Pilot	41	Lung	Mean 66 (SD 10)	29 (71)
Trojan et al [61]	Switzerland	Observational study	6	Prostate, lung, and urothelial	NR	0 (0)
Upton [62]	United Kingdom	Pilot	18	Melanoma	NR	NR
Voruganti et al [63]	Canada	RCT	IG: 24; CG: 24	Breast, colorectal, lung, prostate, ovarian, head and neck, and leukemia, myeloma, or lymphoma	IG: mean 60 (SD 13); CG: mean 60 (SD 14)	IG: 13 (62); CG: 16 (76)
Watanabe et al [64]	Canada	Pilot	44	Breast, lung, and leukemia, myeloma, or lymphoma	Median 60 (range 20-88)	18 (41)
Weaver et al [65]	United Kingdom	Pilot	26	Breast, colorectal	Mean 57	12 (46)
Wright et al [66]	United States	Pilot	10	Gynecologic	Mean 60 (SD 11)	10 (100)
Yanez et al [67]	United States	RCT	74	Prostate	Mean 69 (SD 9)	0 (0)
Yount et al [68]	United States	RCT	IG: 123; CG: 130	Lung	IG: mean 61 (SD 10); CG: mean 60 (SD 10)	IG: 66 (54); CG: 62 (48)

[a]RCT: randomized controlled trial.

[b]IG: intervention group.

[c]CG: control group.

[d]NR: not reported.

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Table 2. Intervention details and engagement outcomes (N=39).

Study	Intervention intensity (duration of the intervention)	Intervention description (content, mode of delivery, health care provider)	Engagement outcomes (frequency, amount, duration, depth, and actual patient engagement)
Alter et al [29]	• Four 30-minute telephone sessions (2 months)	• Content: nurse gathered information on medical and psychological history and discussed effects of cancer on their lives and relationships. Concerns were identified and discussed, strengths in dealing with problems were also identified, and patients were encouraged to use strategies and resources that had been highlighted. • Mode of delivery: telephone. Individual basis. • Health care provider: nurse.	• Frequency: all 4 patients completed all 4 telephone sessions. • Actual patient engagement: 100%.
Badr et al [30]	• Six 60-minute telephone sessions (6 weeks)	• Content: a manual was used covering six areas: self-care, stress and coping, symptom management, effective communication, problem solving, and maintaining and enhancing relationships. Telephone sessions reviewed the content of the manual with patients and carers and set homework for following week. • Mode of delivery: telephone. Patient–caregiver dyads. • Health care provider: trained therapist in mental health counseling.	• Frequency: 90% of patient–caregiver dyad phone calls were made on time. One member had scheduling conflicts, but all were made up with another call. • Actual patient engagement: 100%.
Basch et al [31]	• Participants remained in the study until treatment had concluded or they had died. All intervention participants reported symptoms on tablet or computer kiosks at clinic, but computer-literate participants also sent weekly emails to complete surveys at home (not set).	• Content: participants who were computer-experienced completed symptom-tracking surveys in between clinic visits; if symptoms worsened, this would trigger an email alert to nurses, and participants were encouraged to call if concerned. Those who were computer-inexperienced completed surveys at the clinic before meeting with their clinician. Reports were provided to clinicians but no guidance on what action to take. • Mode of delivery: computer or tablet. Individual basis. • Health care provider: nurses and oncologists.	• Frequency: 73% of intervention participants completed a symptom self-report at any clinic visit, but this did not lead to a difference in the number of nurse calls received compared with the control group (12.8 vs 12.9).
Bensink et al [32]	• Individually tailored. No set engagement (not set).	• Content: the families were provided with videoconference technology, which was used to provide patient assessment and monitoring, family education, communication, and counseling by nurses and other support by social workers or other medical staff. • Mode of delivery: teleconference. Individual basis. • Health care provider: nurses and social workers.	• Frequency: 7 of 11 families received telephone calls, with a total of 25 made and an average of 2.3. • Amount: calls lasted for a median length of 20 (IQR 15-33) minutes.
Bouchard et al [33]	• Ten 90-minute group sessions (10 weeks)	• Content: involved group teleconferences teaching stress and self-management skills for men with prostate cancer with disease-relevant examples. • Mode of delivery: teleconference and telephone. Group delivery. • Health care provider: therapist.	• Frequency: an average of 7.5 (SD 3.1) sessions were attended for the intervention group. • Actual patient engagement: 75%.

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Study	Intervention intensity (duration of the intervention)	Intervention description (content, mode of delivery, health care provider)	Engagement outcomes (frequency, amount, duration, depth, and actual patient engagement)
Bruera et al [34]	• 4-6 calls (2 weeks)	• Content: the calls involved symptom assessment, a review of the types and dosages of medications and their effects, and psychosocial support and patient education. The patient could ask questions, and the nurse asked about their well-being. • Mode of delivery: telephone. Individual basis. • Health care provider: nurse.	• Frequency: no significant difference in the number of phone calls received across any of the four groups: drug and intervention phone call (median 5, IQR 4-6), drug and control call (median 4, IQR 3-5), placebo and intervention phone call (median 5, IQR 4-6), and placebo and control call (median 4, IQR 4-5).
Chambers et al [35]	• Eight 75-minute group sessions (8 weeks)	• Content: an introductory call was used to prepare participants for the group call, and a workbook was used to also guide these group calls. The group calls encouraged peer interaction to support learning mindfulness skills and tackling challenges. Participants were encouraged to engage in 1 mindfulness meditation daily. • Mode of delivery: teleconference. Group delivery. • Health care provider: health professional.	• Frequency: 28% (n=26) attended 0 sessions, 20% (n=19) attended 1 to 3 sessions, 22% (n=21) attended 4 to 7 sessions, and 30% (n=28) attended 8 sessions. • Amount: the average length of a session was 85 (SD 12) minutes.
Chavarri-Guerra et al [36]	• Individually tailored. No set engagement (not set).	• Content: care needs assessments were administered remotely; the multidisciplinary team met to discuss intervention plans, which were then put to the patient. If acceptable, these were then conducted remotely. • Mode of delivery: teleconference, telephone, and SMS text messaging. Individual basis. • Health care provider: multidisciplinary team.	• Frequency: 163 supportive care interventions were provided to 45 patients (median number of interventions per patient 3, range 1-13). • Amount: 0-15 minutes: 38 (23.3%), 16-30 minutes: 58 (35.6%), 31-45 minutes: 37 (22.7%), >45 minutes: 29 (17.8%), (SMS text messaging): 1 (0.6%). • Depth: psychological care: 54 (33.1%), pain and symptom control: 41 (25.1%), nutritional counseling: 20 (12.6%), physical therapy: 14 (8.5%), end-of-life care: 13 (7.9%), geriatric assessment: 8 (4.9%), advance directive completion: 8 (4.9%), psychiatric care: 5 (3%).
Cheung et al [37]	• Five 1-hour sessions (5 weeks)	• Content: each session taught participants 3 out of 8 skills (noticing positive events, capitalizing on or savoring positive events, gratitude, mindfulness, positive reappraisal, focusing on personal strengths, setting and working toward attainable goals, and small acts of kindness), and they were instructed to practice every day. • Mode of delivery: web-based. Individual basis. • Health care provider: unclear.	• Frequency: all 12 participants completed 1 session, 11 participants completed 2 sessions, and 10 participants completed all 5 sessions.
Cheville et al [38,39]	• 8 telephone sessions with fitness care manager, 8 sessions with PT[a] (more if PT thought needed), and pain management intervention arm received call from pain care manager, who then monitored patient-reported pain levels over the course of the study (4 weeks)		

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Study	Intervention intensity (duration of the intervention)	Intervention description (content, mode of delivery, health care provider)	Engagement outcomes (frequency, amount, duration, depth, and actual patient engagement)
		• Content: intervention group 1: tele-delivery of rehabilitation services. Education on role of physical activity in symptom management, consequences of cancer and cancer treatment on loss of muscle bulk and power, and adverse symptoms during exercise. REST[b] to improve functional status. FSP[c] to increase activity levels and aerobic conditioning. Treatment of physical impairments (if any detected) through PT treatment plans. Intervention group 2: same as group 1 with additional pain management to monitor and adjust dosages and medication as needed. • Mode of delivery: telephone and in person. Individual basis. • Health care provider: primary care team, a PT acting as a fitness care manager, a physical medicine and rehabilitation physician, and a local physical therapist.	• Frequency: no difference in remote monitoring contacts across the three groups: mean 10.3 (SD 4.4), mean 10.7 (SD 5.2), and mean 10.2 (SD 4.5). Contacts with the fitness care manager were similar across IG[d] 1 and 2 (mean 7.6, SD 2.9, range 1-21 vs mean 7.2, SD 3.1, range 1-22). The proportion of surveys completed via the web as opposed to the IVR[e] surveys was similar for each arm: CG[f]: 1648 (66%), IG 1: 1721 (74%), and IG 2: 1632 (69%). • Amount: time spent with the fitness care manager was also similar across IG 1 and 2: mean 16.2 (SD 15.2, range 1-124) minutes for IG 1 and mean 16.6 (SD 15.4, range 1-87) minutes for IG 2. • Actual patient engagement: IG 1: 95%; IG 2: 90%.
Chow et al [40]	• 5 telephone sessions (12 weeks)	• Content: patients completed surveys on symptom distress, any questions were referred to palliative nurses, and clinic visits were only scheduled when necessary. • Mode of delivery: telephone. Individual basis. • Health care provider: health care professional trainee.	• Frequency: of the 190 patients, 62% completed the week 1 and 2 phone call, 57% completed the week 4 phone call, 44% completed the week 8 phone call, and 40% completed the week 12 phone call. • Actual patient engagement: 53%.
Cluver et al [41]	• Six 60-minute sessions (not reported)	• Content: sessions involved cognitive therapy. • Mode of delivery: telephone and in person. Individual basis. • Health care provider: therapist.	• Frequency: of the 53 completed sessions, 21 were conducted via videophone, and 32 were conducted face-to-face. One session was missed.
Dixon et al [42]	• 2 telephone sessions (4 weeks)	• Content: follow-up calls following radiation therapy were used to monitor patients' symptoms. • Mode of delivery: telephone. Individual basis. • Health care provider: radiation therapist.	• Frequency: 72% (38/53) of patients completed the telephone assessment at the 1- or 4-week intervals. • Actual patient engagement: 72%.
Donovan et al [43]	• Based upon participants' engagement (3 weeks)	• Content: patients had 3 target symptoms that they worked with the nurse to manage through the message board. The intervention encouraged the patient to understand their problem, discuss their concerns, and understand that they could make positive changes to manage their symptoms. Gaps in knowledge were addressed, and the benefits of new strategies were discussed as well as the setting of goals to achieve these. The patient was then followed up to see whether this worked or whether modifications needed to be made. • Mode of delivery: web-based. Individual basis. • Health care provider: nurse.	• Frequency: the mean number of postings for the 33 women randomized into WRITE[g] Symptoms was 15.87 (median 14, range 0-41). • Amount: the mean length of participant posts was 260.50 (median 210, range 0-808) words. • Duration: for those completing the intervention, it took the nurse–participant dyads an average of 79 (median 76, range 37-185) days to complete all elements of the intervention. • Depth: 25 (75.8%) participants assigned to WRITE Symptoms completed all elements of the intervention.
Eldeib et al [44]	• Weekly calls (dependent on length of treatment)		

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Study	Intervention intensity (duration of the intervention)	Intervention description (content, mode of delivery, health care provider)	Engagement outcomes (frequency, amount, duration, depth, and actual patient engagement)
		• Content: phone calls were used to assess any adverse effects and recommend suitable strategies to remedy this. Adherence to medication was also reinforced. • Mode of delivery: telephone. Individual basis. • Health care provider: pharmacist.	• Amount: total duration of calls was 1554 minutes; average of 35.3 minutes per patient (n=44).
Flannery et al [45]	• 8 telephone sessions (8 weeks)	• Content: nurses phoned participants weekly and assessed their symptoms on 16 common symptoms experienced by those with lung cancer. Any reported symptom required asking questions about the somatic aspects of the symptom. • Mode of delivery: telephone. Individual basis. • Health care provider: nurse.	• Frequency: of the 57% (17/30) of participants retained in the intervention arm, the mean number of intervention calls received was 5.50 (SD 2.48); 8 of 17 participants received all 8 interventions. • Actual patient engagement: 68.8%.
Fleisher et al [46]	• Dependent on participant engagement with web-based survey and skills module (not reported)	• Content: a web-based survey on patient goals, values, and communication preferences, followed by a training module on communication skills. A report was generated for the physician to help guide their next session. • Mode of delivery: web-based. Individual basis. • Health care provider: oncologist.	• Frequency: 18 began the communication aid, and 15 completed it. • Amount: the average time for completing the entire program was 65 minutes—52 minutes spent on the survey and 13 spent on the module. • Actual patient engagement: 83.3%.
Fox et al [48]	• Ten 90-minute sessions (10 weeks)	• Content: facilitator-led relaxation exercises (eg, deep breathing, progressive muscle relaxation, mindfulness meditation, and guided imagery). Psychoeducational sessions focused on stress management. Participants also given homework to practice skills learned in weekly sessions. • Mode of delivery: web-based. Group delivery. • Health care provider: therapist.	• Frequency: week 1: 74% (n=70) attended IG meeting, and 75% (n=73) attended CG meeting. Week 10: 73% (n=69) attended IG meeting, and 82% (n=80) attended the CG meeting.
Fox [47]	• 1 telephone call (not set)	• Content: the outreach call was tailored to the needs of the participant and considered their internal and external environments, including mental, physical, spiritual, psychological, cognitive, relational, social, and cultural aspects. • Mode of delivery: telephone. Individual basis. • Health care provider: social worker or counselor and nurse.	• Amount: mean duration of calls was 56.5 (SD 15.72) minutes. Approximately 71% of calls lasted ≤1 hour.
Friis et al [49]	• Once a week for 4-week web-based symptom reporting, telephone call if threshold exceeded (4 weeks)	• Content: patients filled in health questionnaires in real time, which could be accessed by their health team. Those who needed clinical attention had alerts sent to the clinical team. • Mode of delivery: web-based and telephone. Individual basis. • Health care provider: nurse.	• Frequency: 55% (37/67) of questionnaires answered exceeded the threshold and led to further action by a clinical nurse. Approximately 30% (20/67) of the questionnaires resulted in a phone call.
Gustafson et al [50]	• Dependent on participant engagement (25 months long or 13 months after patient death for caregiver)		

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Study	Intervention intensity (duration of the intervention)	Intervention description (content, mode of delivery, health care provider)	Engagement outcomes (frequency, amount, duration, depth, and actual patient engagement)
		• Content: access to Coping with Lung Cancer website, which provided information on lung cancer, care giving, and bereavement. It also acted as a communication channel between peers, experts, and clinicians. Feedback was also provided by algorithms based on collected data. Tools to help organize support were also provided. Clinicians received reports before next clinic appointments as well as email alerts when high symptom ratings were reported. • Mode of delivery: web-based. Patient–caregiver dyad. • Health care provider: oncologist and enrollment coordinator.	• Frequency: CHESS[h] was used at least once by 73.4% of caregivers and 50% of patients, and 51.6% of caregivers and 34.7% of patients used CHESS ≥5 times. • Amount: the median number of minutes of CHESS use was 103 for caregivers and 146 for patients. • Depth: the median number of pages viewed was 147 for caregivers and 243 for patients.
Haddad et al [51]	• 2 telephone sessions (4 weeks)	• Content: participants were asked about their symptoms, side effects, and drug dosage. • Mode of delivery: telephone. Individual basis. • Health care provider: nurse and radiation therapist.	• Frequency: successful contact at week 1 and 4 was achieved for 22 participants of group A, 14 participants only contacted at week 1, and 3 participants only contacted at week 4. A total of 17 participants were not contacted. • Actual patient engagement: 54.5%.
Hennemann-Krause et al [52]	• Web conferences weekly and face-to-face meetings monthly (continued until patient death)	• Content: symptoms were assessed on a scale, and complaints from patients were listened to. In videoconferences, discrepancy between what the patients reported and what the physician could see on-screen were evaluated. • Mode of delivery: teleconference, email, telephone, and in person. Individual basis. • Health care provider: physicians, nurse, social worker, psychologist, and music therapist.	• Frequency: in-person consultations: mean 7.42 (SD 6.29), web conferences: mean 6.42 (SD 7.64), and total contacts: mean 25.4 (SD 16.3). • Duration: the mean monitoring time was 195 (SD 175.1) days.
Keikes et al [53]	• 2 face-to-face consultations and web-based access to decision support tool in between meetings (not reported)	• Content: treatment options were discussed with oncologist, and the patient reviewed information available on the web and completed questions on treatment goals. • Mode of delivery: web-based. Individual basis. • Health care provider: oncologist and a helpdesk.	• Frequency: 301 patients received a consultation sheet, of whom 155 patients participated in the web-based part of the decision tool (51%). • Amount: the median overall time spent on web-based decision support was 38 (IQR 18-56) minutes. Time spent was highest on reading treatment background information (median 4, IQR 1-11 minutes) and answering questions about patients' perspective (median 5, IQR 2-11 minutes). • Actual patient engagement: 51%.
Liu et al [54]	• Twice daily reporting of blood pressure and diarrhea data reported as needed. Algorithmic feedback and prompts to call HCP[i] when appropriate (4 weeks).	• Content: participants reported blood pressure and diarrhea entries, which triggered algorithmic feedback, and the clinical team reviewed this. Email alerts were sent to the clinical team for high results or when a blood pressure check was missed. • Mode of delivery: mobile app. Individual basis. • Health care provider: patients' clinical team.	• Frequency: patients using eCO[j] recorded 98.2% of expected home blood pressure values. All 12 patients were prompted to call at least once, with most being prompted 7 to 20 times. One patient was prompted 54 times but was considered noncompliant. • Actual patient engagement: 98.2%.

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Study	Intervention intensity (duration of the intervention)	Intervention description (content, mode of delivery, health care provider)	Engagement outcomes (frequency, amount, duration, depth, and actual patient engagement)
Nemecek et al [55]	• Participant-dependent reporting and contact with physician (until participant death)	• Content: VSee was used to connect patients and their physicians when the patient required medical advice. This was available around the clock. Patients could also input vital signs (temperature, blood pressure, pulse, and oxygen saturation) as well as treatment and other variables (pain, nutrition, and body weight). This could then be reviewed by the physician in charge. • Mode of delivery: teleconference. Individual basis. • Health care provider: physician.	• Frequency: a total of 37 telemedical requests were submitted, of which 35 were successful, whereas 2 failed. A total of 638 data entries were performed. Entry count varied between 1 and 265 per patient.
Rasschaert [56]	• Reported daily treatment intake, toxicity, and disease-related symptoms. Calls made when toxicity levels were high (no set duration; patients used for duration of oral anticancer agent).	• Content: participants were asked to self-report disease-related symptoms and treatment toxicity via an app. This could be accessed by physicians and cancer care providers at clinic visits or when admitted to hospital. Alerts would be sent to caregivers or phone calls would be organized when high toxicities were reported, and the participants were also told to seek help. • Mode of delivery: smartphone. Individual basis. • Health care provider: data manager, physician, and other health care professionals.	• Frequency: average daily compliance with registration of treatment intake was 91.2%. • Duration: 5 patients used the coach >4 weeks (and only 1 used it for >12 weeks). • Actual patient engagement: 91.2%.
Rose et al [57]	• 1 face-to-face meeting, 1 follow-up call. Patients could then contact the nurse 24 hours a day, 7 days a week at their convenience (2 months).	• Content: the initial meeting occurred in the patient's home and was to set goals for patient communications and shared decision-making. Coping and communication issues, strategies to address problems, and concerns and expectations were also discussed. Follow-up calls covered the multifaceted impact of cancer and treatment, preparing patients for future therapy or progression, identifying goals either personal or of treatment, identifying further needs of support, supporting positive emotions of oneself, encouraging independence and coping, optimizing social support, addressing practical problems, and referring patients for additional support. • Mode of delivery: telephone, email, or in person. Individual basis. • Health care provider: nurse.	• Frequency: average number of monthly contacts was higher among middle-aged group (mean 2.6, SD 2.7) than among the older age group (mean 2.0, SD 1.2). • Amount: average length of calls was 10-11 minutes. • Duration: average of 62 days of access to intervention.
Sardell et al [58]	• 3 monthly telephone calls and 1 face-to-face clinic visit at the fourth month. Telephone calls continued if no recurrent or progressive disease (4 months but also participant-dependent).	• Content: the telephone calls followed a semistructured script, which allowed patients to talk freely about their symptoms, how they were feeling, and any problems they had. More structured questions on their neurological status, medication, use of hospital services, return to work, and social activities followed. • Mode of delivery: telephone. Individual basis. • Health care provider: nurse.	• Frequency: a total of 254 telephone calls were made, with a median of 4 calls per patient (range 1-14). • Amount: median time on calls was 10 (range 2-10) minutes. • Duration: median time was 6 (range 2-21) months.
Schmitz et al [59]			• Duration: average use of the tablet was 69.9 days for 7 participants.

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Study	Intervention intensity (duration of the intervention)	Intervention description (content, mode of delivery, health care provider)	Engagement outcomes (frequency, amount, duration, depth, and actual patient engagement)
	• Daily app notifications to engage and 1 weekly phone call with navigator (12 weeks)	• Content: participants received a daily prompt to interact with the app. The app asked a symptom question, which, when answered, prompted different facial expressions from the nurse avatar and different verbal responses. Navigator calls focused on reviewing symptoms and steps, which were compiled in a report and emailed to the clinical care team. • Mode of delivery: mobile app and telephone. Individual basis. • Health care provider: patient navigators.	
Sherry et al [60]	• Pamphlet and 1 telephone session (1-3 days)	• Content: a personalized pamphlet was presented to the patient based upon problems they noted when completing a distress survey. This was followed up by a phone call a couple of days later to answer any questions and to check understanding. The coach offered referrals to social work, palliative and supportive care services, physical therapy, integrative medicine, financial services, and nutrition. • Mode of delivery: telephone and in person. Individual basis. • Health care provider: nurse.	• Frequency: all patients reported that they had read the education pamphlet and received the coaching call.
Trojan et al [61]	• Participant-dependent reporting of symptoms and side effects (3 months)	• Content: patients reported the number, characteristics, and intensity of symptoms and therapy side effects. The symptom severity could trigger alerts to the on-call oncologist, which could result in a telephone consultation. • Mode of delivery: mobile app and telephone. Individual basis. • Health care provider: oncologist.	• Frequency: 1279 symptom entries were recorded. Number of symptom data entries from the 6 patients ranged from 31 to 458 within the 3-month period. A total of 4 of the 6 patients also triggered 14 alerts, all of which correlated to cough, respiratory stress, fever, and fatigue and made patients aware of making contact with their treating center. A total of 6 alerts resulted in telephone consultations with the treating center or oncologist on call.
Upton [62]	• 1 telephone assessment (1 day)	• Content: before ipilimumab infusion, the patient's blood was tested, and immune-related adverse events were assessed by the nurse. After the infusion, patients were contacted weekly to monitor for immune-related adverse events and for the nurse to provide advice. Patients were also asked to call a 24-hour triage service if experiencing any problems. • Mode of delivery: telephone and in person. Individual basis. • Health care provider: nurse.	• Frequency: over a 1-year period, a total of 56 telephone assessments were undertaken.
Voruganti et al [63]	• Dependent on participant engagement with web-based messaging and communication with HCPs (not reported)		• Frequency: over the study period, most (17/20, 85%) Loops (web-based tool to facilitate communication) had message exchanges, with 65% (13/20) having >6 messages exchanged. During the study, there were 358 log-ins by all participants: 43 on the mobile version and 315 on the desktop version.

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Study	Intervention intensity (duration of the intervention)	Intervention description (content, mode of delivery, health care provider)	Engagement outcomes (frequency, amount, duration, depth, and actual patient engagement)
		• Content: the web-based communication tool (Loop) facilitated conversations between patients, caregivers, and health care providers. There was no set communication the tool should be used for, only that it should not be used for urgent communication. • Mode of delivery: web-based. Individual basis. • Health care provider: oncologist, palliative care physician, and other health care professionals.	
Watanabe et al [64]	• One 90-minute videoconference with a 30-minute follow-up if necessary (1 day)	• Content: patients arranged to attend a local clinic, where a videoconference could be set up with the cancer institute. Blood tests, radiological investigations, and patients' symptoms and needs were assessed before this, and the results were shared with the team. A total of 3 team members, including the physician, could be on the videoconference, with every member given 15 minutes to interview the patient. After the assessments, the team formed a management plan in discussion with the patient and family, which was sent to the patient's GP[k]. • Mode of delivery: teleconference. Individual basis. • Health care provider: nurses, dieticians, psychologists, respiratory therapists, social workers, occupational therapists, physical therapists, speech language pathologists, radiation oncologists, and pharmacists.	• Frequency: a total of 72 clinic visits took place, consisting of 44 initial consultations and 28 follow-up visits. • Depth: variety of members of MDT[l] seen at consultations: dieticians (56.8%), psychologists (27.3%), respiratory therapists (15.9%), social workers (13.6%), occupational therapists (9.1%), physical therapists (9.1%), and speech language pathologists (4.5%). • Actual patient engagement: 100%.
Weaver et al [65]	• Phone app used twice daily to report symptoms; alerts to nurse generated if toxicity was high or the patient had not self-reported for a while (while on treatment)	• Content: patients asked to fill out a short diary containing entries for temperature, diarrhea and assessments for vomiting, nausea, mucositis, hand–foot syndrome, and—for patients receiving oxaliplatin—peripheral neuropathy. Alerts were triggered based upon toxic side effects or a lack of reporting, with a nurse available to provide clinical advice. • Mode of delivery: mobile app. Individual basis. • Health care provider: nurse.	• Frequency: the patients completed the diary on 92.6% of occasions (range 73.7%-100%). On 396 occasions, self-care advice messages were sent to the patients. • Actual patient engagement: 92.6%.
Wright et al [66]	• Daily app notifications for 30 days. If high-risk symptoms were reported, the patient was told to contact the clinician (30 days).	• Content: participants completed daily surveys on quality of life, physical function, and symptoms, of which they ranked the severity. High-risk symptoms initiated a prompt to contact the participant's clinician with an in-built call button. • Mode of delivery: mobile app and telephone. Individual basis. • Health care provider: oncologists and researchers.	• Frequency: study participants were 70% adherent to smartphone surveys. A total of 7 participants answered daily surveys ≥4 times a week. • Actual patient engagement: 70%.
Yanez et al [67]	• Ten 90-minute group sessions (10 weeks)		

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Study	Intervention intensity (duration of the intervention)	Intervention description (content, mode of delivery, health care provider)	Engagement outcomes (frequency, amount, duration, depth, and actual patient engagement)
		• Content: participants were taught a stress reduction or relaxation technique while also developing stress awareness, learning stress reduction skills, changing negative stressor appraisals, developing coping skills, building interpersonal skills, and building or enhancing social networks. They were also encouraged to access the website, which contained material related to each group session and videos to review in between sessions. • Mode of delivery: teleconference. Group delivery. • Health care provider: therapists.	• Frequency: HP[m] participants completed significantly more sessions (mean 8.22, SD 2.75 compared with mean 6.59, SD 3.72) than CBSM[n] participants. HP participants also completed significantly more weekly assessments (mean 7.05, SD 3.14) vs mean 4.84, SD 3.35) compared with the CBSM condition. • Actual patient engagement: 65.9%.
Yount et al [68]	• Weekly calls to report symptoms, alerts triggered calls from a nurse (12 weeks)	• Content: participants completed a symptom survey over the phone using the telephone keypad. Clinically significant symptoms were automatically reported to the clinical team for assessment and management with a nurse phone call. Data were also provided to physicians every 3 weeks before visits to facilitate discussion. • Mode of delivery: telephone. Individual basis. • Health care provider: physicians.	• Frequency: compliance with completion of weekly symptom monitoring phone calls was 82.1%. • Actual patient engagement: 80.8%.

[a]PT: physical therapist.
[b]REST: Rapid Easy Strength Training.
[c]FSP: First Step Program.
[d]IG: intervention group.
[e]IVR: interactive voice response.
[f]CG: control group.
[g]WRITE: Written Representational Intervention To Ease Symptoms.
[h]CHESS: Comprehensive Health Enhancement Support System.
[i]HCP: health care professional.
[j]eCO: eCediranib/Olaparib.
[k]GP: general practitioner.
[l]MDT: multidisciplinary team.
[m]HP: health promotion.
[n]CBSM: cognitive behavioral stress management.

Engagement

The engagement outcomes for all studies are outlined in Table 2.

Frequency

Across most studies (36/39, 92%), the frequency of times contact was made with the intervention was reported [29-43,45-47,49-58,60-68]. There was substantial heterogeneity in the measurement of frequency across studies. Of the 39 studies, 13 (33%) reported the percentage of contacts either with the whole intervention or with each individual intended session [30,31,35,40,48-50,54,56,63,65,66,68]. The number of contacts with the intervention overall or each individual session was reported by 69% (27/39) of the studies [29,33,34,36-39,41-43,45,46,49,51-55,57,58,60-67].

Across 44% (17/39) of studies, it was possible to create a standardized percentage of actual patient engagement compared with intended engagement [29,30,32,33,38-40,42,45,46,51,53,54,56,63,66-68]. This ranged from 51% [53] to 100% [29,30,64], with an average across all 17 studies of 75.4% (SD 15.8%). In the remaining 49% (19/39) of studies, it was not possible to create this standardized statistic because of a lack of reported data, and the design of the intervention meant there was no intended engagement and it was instead tailored to the patients' needs.

Amount

A total of 31% (12/39) of studies measured the amount of contact with each intervention or with the intervention overall [32,35,36,38,39,43,44,46,47,50,53,57,58]. Of the 39 studies, 3 (8%) measured the average amount of time of each intervention

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contact (10.5 to 85 minutes) [35,47,57], 2 (5%) reported the average amount of time across all intervention contacts (16 to 65 minutes) [38,39,46] and 1 (3%) reported the total amount of call durations, which could be averaged across all intervention participants to 35.3 minutes [44]. In total, 5% (2/39) of studies reported the median amount of time for each intervention contact (10 to 20 minutes) [32,58], and 5% (2/39) of studies reported the median amount of time across the whole intervention (38 to 146 minutes) [50,53]. A total of 3% (1/39) of studies reported the number of intervention contacts that fell into a range of minutes (eg, 16-30 minutes: 58 contacts) [36]. A total of 3% (1/39) of studies did not report time but, as it was a web-based intervention with communication with the health professional through posts on a message board, instead reported the average length of each post at 260.5 words [43].

Duration

A total of 15% (6/39) of studies that had open-ended interventions reported the length of time that each participant was exposed to the intervention [43,52,56-59]. A total of 10% (4/39) of studies reported the average time of exposure to the intervention, ranging from 62 to 195 days [43,52,57,59]. A total of 3% (1/39) of studies reported a median amount of exposure to the intervention of 6 months [58], and the final study (1/39, 3%) reported the number of participants exposed for >4 weeks (n=5) and >12 weeks (n=1) [56].

Depth

A total of 10% (4/39) of studies reported on the variety of components of the intervention that the participants accessed [36,43,50,64]. Each study measured depth in different ways. A total of 3% (1/39) of studies reported the percentage of time that each health professional was on the teleconference calls [64], and another study (1/39, 3%) simply reported that 75% of patients had completed all elements [43]. The number of different interventions that all participants received was reported by 3% (1/39) of studies [36], and the final study (1/39, 3%) reported that patients had viewed a median of 243 webpages [50].

Association With Intervention Level of Intensity

Expected levels of engagement for both patients and health professionals were reported across low (≤3 contacts), medium (4-7 contacts), and high (≥8 contacts) categories. A total of 13% (5/39) of studies could not be categorized as there was no expected engagement with the intervention, and the extent of engagement was determined at the patient's discretion [32,36,43,55,63]. Table 3 shows the number of studies with the expected interaction of both the patient and health professional with the intervention. Most studies expected a similar level of interaction from both the patient and health professional in an intervention, but no studies expected more interaction from the health professional than from the patient.

Table 3. Number of studies with the expected engagement of the patient and health professional (n=34).

Expected patient interaction with the intervention	Expected health professional interaction with the intervention		
	Low, n (%)	Medium, n (%)	High, n (%)
Low	10 (26)	__[a]	—
Medium	2 (5)	8 (21)	—
High	7 (18)	—	7 (18)

[a]No data available for category.

Figures 2 and 3 are graphical representations of the association between expected levels of engagement for the patient (Figure 2) and the health professional (Figure 3) and the percentage of actual engagement with the intervention by the patient. Figure 2 shows that the studies that had low expected engagement for the patients had a combined actual patient engagement of 64% (SD 14.8%); for medium expected engagement, this was 66.9% (SD 16.4%); and, for high expected engagement, this was 87% (SD 8.2%). Figure 3 shows that the category with the highest level of combined actual patient engagement was the studies that expected the health professionals to have a high level of engagement with the intervention (86.6%, SD 8.3%). The studies in the categories of low and medium expected engagement from health professionals had lower levels of combined actual patient engagement (71%, SD 15.2% and 62.3%, SD 15%, respectively).

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Figure 2. Box plot to present the association between expected levels of engagement by the patient and the percentage of actual engagement by the patient.

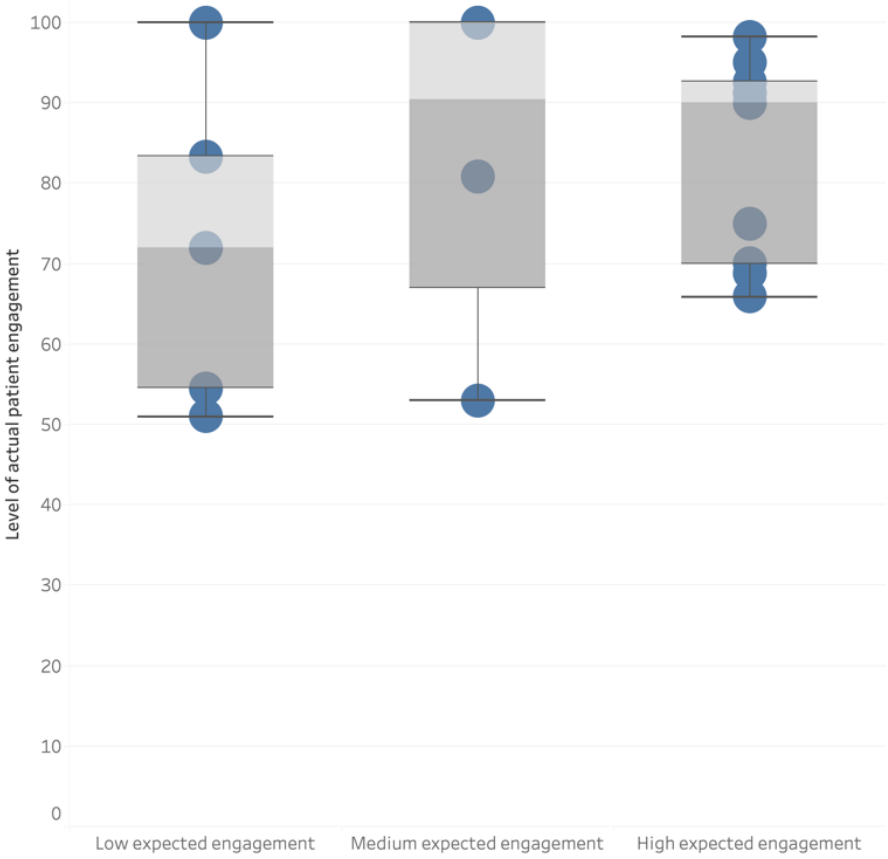
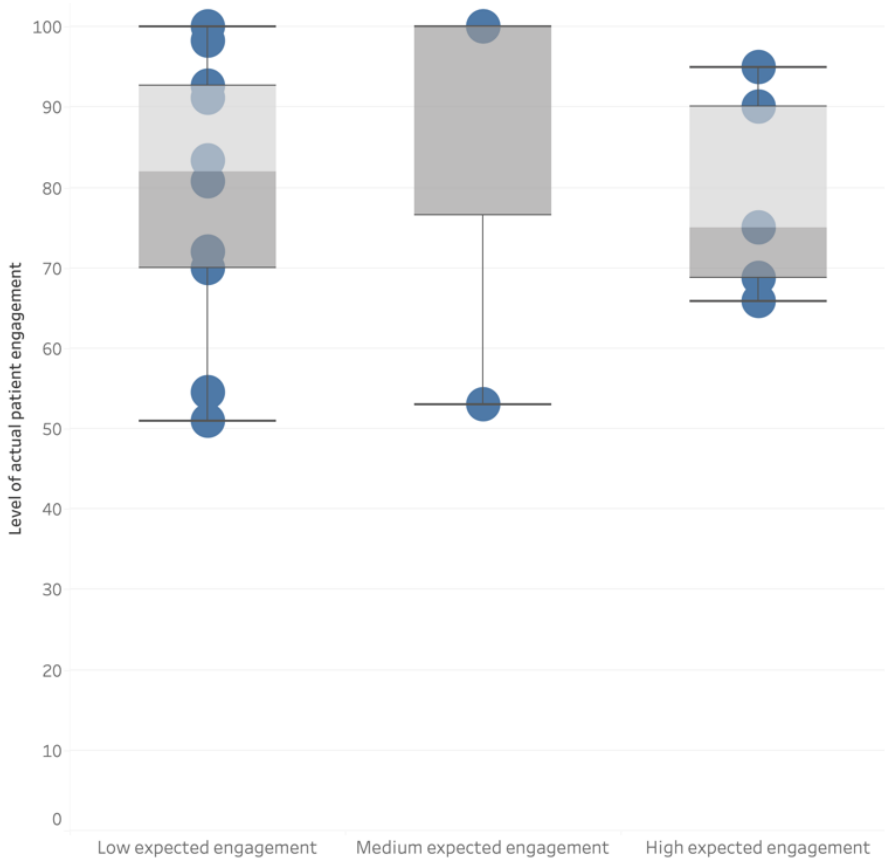


Figure 3. Box plot to present the association between expected levels of engagement by the health professional and the percentage of actual engagement by the patient.



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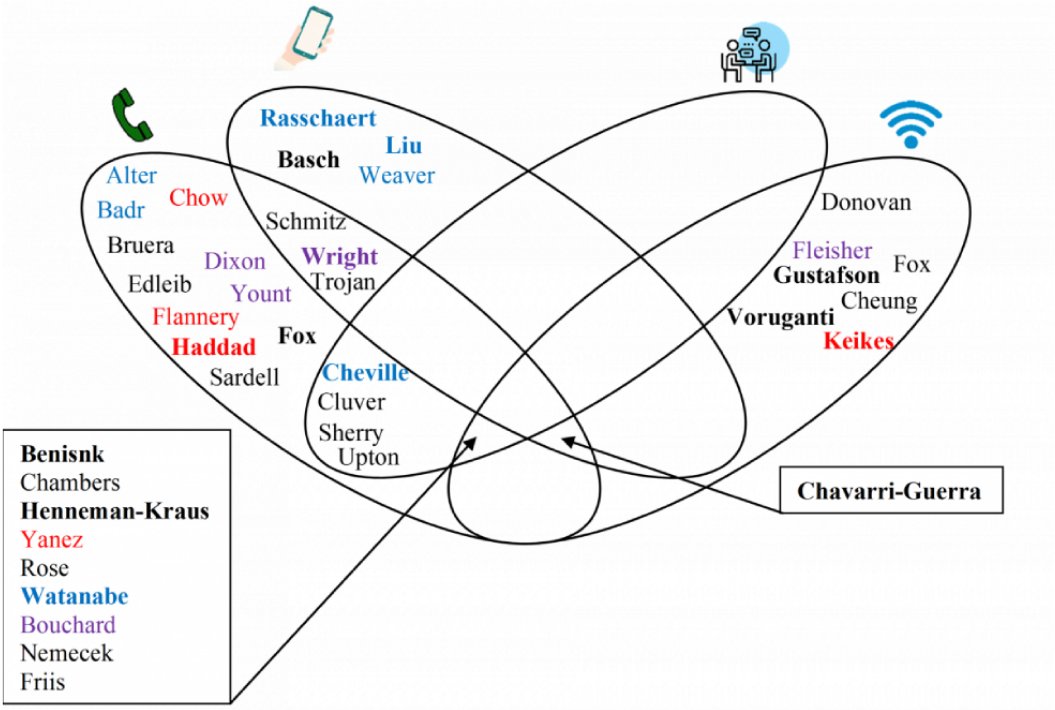
Association With Intervention Mode of Delivery and Health Care Providers

Figure 4 [29-68] shows the modes of delivery of each intervention and where interventions use multiple modes, with the names in bold involving multiple health professionals. The figure also shows, where available, the percentage of actual patient engagement by way of color, with blue showing 90% to 100%, purple showing 70% to 89%, and red showing <70%. Of the 39 studies, 17 (44%) used multiple modes of delivery, whereas the remaining 22 (56%) used 1 mode. The telephone was the most popular mode of delivery (28/39, 72%) followed by web-based delivery of the intervention (17/39, 44%). The use of only a tablet or smartphone app for the intervention appeared to be associated with the most actual patient engagement with an intervention, with 8% (3/39) of studies showing between 90% and 100% engagement [54,56,65]. The use of a telephone was more mixed, with actual patient engagement ranging from 54.5% [51] to 100% [29,30]. Figure 4 also shows broadly how many health care providers were involved in delivering the interventions, with those involving multiple health care providers shown in bold. Those interventions that involved multiple health care providers reported higher patient engagement than those with only 1 health care provider (79.3%, SD 18.5% vs 70.5%, SD 11.5%).

Figure 4. Modes of delivery of each intervention and, where reported, the percentage of actual frequency of engagement [29-68].



Note: From left to right each oval represents: left – Interventions conducted by telephone; middle-left – interventions conducted via smartphones, tablets and mobile apps; middle-right – interventions conducted face-to-face; right – interventions that are web-based. The **bold** names of the first authors indicate interventions with the involvement of multiple health professionals. The colours refer to the percentage of actual frequency data: blue – 90-100%; purple – 70-<90%; red <70%; black – no data available. The percentage of actual frequency data is calculated from the expected amount of engagement a patient was meant to undertake in the intervention.

Study Quality

The included studies could be grouped into two broad categories to be assessed using the Mixed Methods Appraisal Tool: quantitative RCTs and quantitative nonrandomized trials. The RCTs were of a broadly high quality; however, a number of studies did not provide enough information to assess whether the randomization procedure was conducted adequately or whether the groups at baseline were comparable. There were also 15% (6/39) of studies that did not have complete outcome data at follow-up. Among the nonrandomized trials, study quality was again high, apart from the included studies that did not control for confounders in their analysis. This is likely because most of these studies were feasibility or pilot studies and were not powered to detect significance, which would have been inappropriate. A breakdown of how each study was rated can be found in Multimedia Appendix 2 [29-68].

Discussion

Principal Findings

This systematic review is the first to synthesize engagement data from telehealth interventions for people with advanced cancer. This review found that people with advanced cancer were able to successfully engage in telehealth interventions with variable types of telehealth modalities, including telephone, mobile phone–based apps, and web-based interventions, albeit largely in the context of research studies. This review found that the frequency of engagement with the intervention was the

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most commonly reported measure of engagement, although there was heterogeneity in the method of reporting across the studies. Where standardized comparison was possible across the studies, actual engagement as a proportion of intended engagement was at an average of 75.4% (SD 15.8%). The level of engagement was found to vary based on the expected interaction of both the patient and health care professional and the mode of delivery. Actual patient engagement was higher in studies that expected higher levels of engagement from both the patient and health care professional but was noticeably lower in studies that expected only a low or medium level of engagement. Furthermore, the use of only a tablet or smartphone app for an intervention appeared to be associated with the highest levels of actual patient engagement with an intervention. This could in part be explained by the immediacy of access and reduced steps for accessing an intervention through a mobile phone app when compared with an intervention hosted on a website.

This review is in line with previous reviews that looked at engagement with interventions involving digital technology among people with chronic diseases, which found broadly that there are high levels of engagement with interventions [20]. However, this review provides an overview and critique of existing reporting of engagement for telehealth interventions in patients with advanced cancer and found wide disparities in metrics for engagement used and reported across the included studies. The frequency of interaction with an intervention was reported widely, but other measures of engagement, such as the amount of time spent engaging with the intervention, were not reported as well. Furthermore, the duration and depth of engagement with the intervention were reported by only one-quarter of all included studies (9/39, 23%). This may be due to the design of interventions with a set duration or only 1 component that patients could engage with, but this was not clear across studies. In addition, few studies reported the expected levels of engagement for an intervention, limiting the interpretability of any subsequent reporting of actual patient engagement. Refining and using measures to better understand factors driving digital engagement, including for telehealth, could inform the development of approaches from design through monitoring as part of routine care. For example, the application of engagement measures could serve as a progression criterion in feasibility studies of emerging telehealth approaches. Future research may need to define and develop meaningful and context- and condition-appropriate measures of digital engagement for palliative care to facilitate measurement of digital engagement. Although this review focused on the quantitative measures of behavioral engagement, the future development of a measure should attempt to incorporate components that provide a broader understanding of subjective experiences and aspects of engagement, potentially through qualitative approaches. There is also scope to develop and refine the dimensions comprising the digital engagement framework used to guide the synthesis of data in this study. For example, there is scope to incorporate a temporal element to consider the intensity of the intervention (eg, whether the intervention is spread over a week or months) alongside refining the underpinning definitions of terminology used for each dimension as the framework continues to evolve.

Through this review, we can conclude that there is no standardized method to report engagement in telehealth interventions for people with advanced cancer. The frequency of interactions with the intervention was presented most commonly, although the way in which this was done varied greatly across the studies, and there is a limited ability to understand what this means in the context of the intervention and the proposed and expected engagement needed for clinical utility. For example, people with advanced cancer have fluctuating needs, and a higher level of engagement with an intervention may not relate to the success of the intervention itself but be reflective of worsening outcomes for the patient [69]. In addition, patients may have their symptom management needs met early on in the intervention and may not need further follow-up, which may not be indicative of poor engagement with the intervention per se. With regard to mobile health interventions, the Mobile Health Evaluation, Reporting and Assessment checklist has been developed to help standardize the methodology for reporting the content and context of an intervention to support reproducibility and comparison of interventions [70]. Future iterations of the tool could include, for example, reporting of the expected and actual patient engagement levels of intended users of telehealth interventions alongside frequency of use—the most widely reported measure in this review. These data could complement and contribute to emerging evidence regarding the feasibility and acceptability of telehealth approaches as part of care for people with advanced cancer.

Recent evidence suggests that digital health interventions could provide a degree of efficacy related to QOL and psychosocial well-being [16]. For this review, most included interventions focused on symptom management, with high levels of engagement that suggest potential for its use to support remote monitoring. This approach could facilitate reductions in the required number of in-person visits while enabling continued access to data to inform patient care. However, in order to ensure such an approach is sustainable, there is a need to consider the burden of data entry on patients and the need for review—and potentially response—by health professionals. For patients, emerging approaches provide options for enhancing the richness of data received through remote monitoring without increasing the data burden for patients. For example, wearable technologies can passively collect sensor data on heart rate and activity to inform automatic monitoring and feedback processes [71], augmenting existing approaches without increasing the need for manual data entry. For health professionals, this review found that studies with high levels of intended engagement for both the patient and health care professional were associated with higher levels of actual engagement on the part of the patient. High intended engagement from health care professionals may not be a sustainable approach for digital technology, particularly when considered alongside the additional *invisible* work that such digital health can create for health professionals (eg, data must be interpreted, made sense of, located within existing knowledge and data sets, and negotiated) [72]. This is important to consider in light of projections of an increasing burden of serious health-related suffering and subsequent demands on palliative care services across geographical regions where demand is increasingly

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outstripping supply [73,74]. Therefore, for telehealth approaches to be sustainable as part of care for people with advanced cancer, they should seek to balance demands on both the patient and the care team, seeking to achieve maximal information with minimal data burden.

Limitations

There were a number of limitations associated with this review. First, the focus of engagement in this review was on the behavioral aspects that were outlined by Perski et al [19] but not on the subjective measures of engagement, such as interest, attention, and enjoyment. Integrating these subjective measures into a future mixed methods review could allow us to evaluate the experience of interventions. In addition, because of the heterogeneity of the studies and reported approaches to measuring engagement, such as frequency, it is difficult to determine exactly which components of interventions contribute to higher engagement levels. We were only able to draw associations, and future research is needed to better explore causal factors. Furthermore, although this review looked at the extent of engagement, how it was measured across studies, and the association with the study characteristics, we did not assess whether engagement led to an improvement in patient-reported outcomes or experience. A future review should consider how engagement interacts with patient-reported outcomes. In addition, when determining the categories for low, medium, and high expected engagement, we did not take into account the time frame of the intervention; therefore, 2 studies could be grouped together with different levels of intervention intensity. Furthermore, most of the studies included in this review explored the intervention effect through mostly controlled studies, which could bias the recruitment toward those individuals who were motivated and more likely to be technologically literate. The levels of engagement identified in this review may not then translate into routine clinical care if these studies and their intervention effect have to date been confined to exploration in the context of RCTs and similar study approaches. This review also limited the included studies to those written in English; therefore, this review may not contain the entirety of related studies.

Conclusions

This review identified that, where reported, there is a high level of engagement with telehealth interventions among people with advanced cancer. We identified that actual patient engagement is associated with both the expected level of engagement of the patient and the health professional as well as the mode of delivery of the intervention. We highlighted the heterogeneity in the reporting of engagement results across the research and the need to improve such reporting guidelines. As treatment delivery becomes increasingly more dependent on remote or telehealth modalities, the inclusion of a measure of engagement in future telehealth evaluations is essential to enable the comparisons of interaction and use across intervention approaches and to provide further granularity in factors that determine optimal implementation of telehealth approaches. There is a need for consistent measurement and reporting of domains relating to digital engagement (eg, breadth, duration, and frequency) with the scope to amend or develop measures. This will increase the ease of reporting of engagement in future studies, inform which telehealth intervention components are linked to variations in engagement, facilitate evidence syntheses, and support the development of condition-specific benchmarks of digital engagement for people with advanced cancer.

Acknowledgments

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Conflicts of Interest

None declared.

Multimedia Appendix 1
Search strategy for Ovid MEDLINE.
[DOCX File , 18 KB - cancer_v8i1e33355_app1.docx]

Multimedia Appendix 2
Quality appraisal tables of quantitative randomized controlled trials and nonrandomized trials.
[DOCX File , 20 KB - cancer_v8i1e33355_app2.docx]

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Abbreviations

PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses
PROSPERO: International Prospective Register of Systematic Reviews
QOL: quality of life
RCT: randomized controlled trial

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Original Paper

Assessment of the Quality, Understandability, and Reliability of YouTube Videos as a Source of Information on Basal Cell Carcinoma: Web-Based Analysis

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Abstract

Background: Patients with skin cancer increasingly watch online videos to acquire disease-related information. Until now, no scientific evaluation of the quality of videos available for German-speaking patients with basal cell carcinoma (BCC) has been performed.

Objective: In this study, we aimed to identify and evaluate videos about BCC provided on YouTube.

Methods: A video search on YouTube was conducted in July 2020, using German BCC-related keywords (eg, “Basalzellkarzinom,” “Basaliom,” “weißer hautkrebs,” and “heller hautkrebs”). The first three pages (ie, 60 videos) were searched by two independent researchers for each keyword. Two authors evaluated videos that met the predefined eligibility criteria. The quality of the information of the videos was evaluated using the DISCERN tool and the Global Quality Scale (GQS). The understandability and actionability were assessed with the Patient Education Materials Assessment Tool for Audiovisual Materials (PEMAT-A/V). The reliability was assessed with the JAMA (Journal of the American Medical Association) criteria score. Subgroup differences were identified using the Kruskal-Wallis test.

Results: A total of 41 videos were included in the evaluation. The mean assessment scores were as follows: DISCERN, 3.3 (SD 0.80); GQS, 3.8 (SD 1.1); JAMA, 27.74% (SD 22.1%); understandability, 70.8% (SD 13.3%); and actionability, 45.9% (SD 43.7%). These values indicated that the videos were of medium to good quality and had good understandability, low actionability, and poor reliability. The quality of videos provided by health professionals was significantly higher than that of videos provided by laypersons.

Conclusions: Optimization of health-related videos about BCC is desirable. In particular, adaptation to reliability criteria is necessary to support patient education and increase transparency.

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KEYWORDS

basal cell carcinoma; YouTube; videos; patient education; shared decision-making; quality; reliability; internet; information

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Introduction

Cutaneous basal cell carcinoma (BCC) represents the most common malignant tumor type in Central Europe, accounting for more than 80% of all epithelial skin carcinomas [1,2]. These tumors typically occur among fair-skinned individuals and are located most commonly on the head and neck, followed by the trunk and extremities [3]. The incidence of BCC continues to increase each year, with a current annual incidence of approximately 200 cases per 100,000 persons in Germany. However, the actual number is estimated to be much higher because cancer registries only document the first occurrence of BCC, and multiple tumors are not recorded [2,4]. BCC is rarely fatal, and surgical interventions remain to be the gold standard of treatment [1,5,6].

Patients with cancer in Germany commonly prefer to attend physician consultations in order to acquire disease-related information [7]. However, the physician's time for a consultation is usually limited, while patients receive a large amount of medical and treatment-related information. Thus, patients may struggle with understanding all of the information provided and may subsequently feel inadequately informed [8]. While medical consultations and written information remain to be the most important sources of health information for patients, a steadily increasing number of patients are seeking health information on the internet [7,9-11]. YouTube is an open-access video-sharing platform, ranking second among the most-accessed websites worldwide, as it counts 5 billion visits per day and 1 billion hours watched daily [12]. It is increasingly used to disseminate health-related information and has become an easily accessible source for patients to acquire information related to their diseases [13]. The distribution of medical information to such a huge audience offers invaluable opportunities but also challenges, as the quality of unfiltered information posted can be of low scientific quality [14]. Information may even be misleading or harmful, as the credibility of the providers cannot be verified, and quality control of these videos has not yet been established [15-17]. Until now, no scientific evaluation of the quality of videos available for German-speaking patients with BCC has been performed. Therefore, the aim of this study was to identify YouTube videos about BCC and to assess their quality, reliability, usability, and understandability. The results of this study may encourage shared decision-making and be beneficial for both patients and health care providers in order to recommend appropriate videos to their patients.

Methods

Search Strategy

A video search on YouTube was conducted in July 2020, using German BCC-related keywords (eg, "Basalzellkarzinom," "Basaliom," "weißer hautkrebs," and "heller hautkrebs"). The standard search options provided by YouTube were maintained. The first three pages (ie, 60 videos) were searched by two independent researchers for each keyword using Internet Explorer 11 (Microsoft). It has been observed that a significant proportion of users watch videos from only the first three pages. Furthermore, a similar methodology has been used in previous studies related to YouTube videos [18,19].

Eligibility Criteria

To be eligible for evaluation, videos had to meet the following inclusion criteria: (1) contain information referring to BCC, (2) be accessible for free and for all users, and (3) provide information in the German language. Videos were excluded if they were commercials, they did not have sound, they presented only photos, or if the duration was less than one minute. All search results were screened for duplicates, and the predefined eligibility criteria were applied.

Grouping of Videos

Due to the variety of the video providers, the videos were grouped according to their original source into the following categories: layperson, health professional (ie, hospital or practice), educational provider, noncommercial provider or professional society, pharmaceutical company, health portal, and unclassified. For television or news reports, we distinguished whether they were uploaded by the official channel or reuploaded by private providers.

Data Management

The available baseline information (ie, URL, title, name of the provider, video length, and year of upload) of each selected video was documented. Additionally, the numbers of views, likes, and dislikes were extracted. With this information, we calculated the video power index (VPI) to assess the popularity of the videos. The VPI is calculated as follows:

$$\text{VPI} = \text{number of likes} / (\text{number of likes} + \text{number of dislikes}) \times 100$$

The baseline information was extracted to an internally piloted data extraction sheet using Microsoft Excel 2010.

Two reviewers (TS and MH) independently assessed the videos' quality of information, reliability, and understandability. Prior to the assessment, the use of the assessment tools was piloted by independently evaluating the first five videos to discuss potential difficulties and resolve questions.

Quality of Information

The DISCERN tool is commonly used to assess the quality of cancer information and was developed for laypersons [20]. A modified German version of this tool was used in this study, consisting of nine items that were used (1) to review a video's transparency (items 1-6), (2) to review a video's content (items 7 and 8), and (3) to give an intuitive assessment summary (item 9). Items were scored on a 5-point scale ranging from 1 ("criterion is not met at all") to 5 ("criterion is fully met"; Multimedia Appendix 1). Thus, videos that were rated, on average, 4 or higher were considered to be of good quality, those rated from 2 to below 4 were considered medium quality, and those rated less than 2 were considered low quality. A maximum of 45 points could be achieved.

Additionally, the Global Quality Scale (GQS) was used. The GQS includes a 5-point scale ranging from 1 ("low quality") to 5 ("high quality") [21]. Videos scoring 4 or 5 points were rated

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as high quality, those scoring 3 points were rated as medium quality, and those scoring 1 or 2 points were rated as low quality.

Understandability and Actionability

The Patient Education Materials Assessment Tool for Audiovisual Materials (PEMAT-A/V) was chosen to assess the individual videos' understandability and actionability. The understandability section comprises 13 items that covered content, word choice and style, organization, layout and design, and the use of visual aids [22]. The second section covers actionability by four items. Each item can be scored as 0 ("disagree"), 1 ("agree"), or N/A ("not applicable"). Then, percentage scores for both sections are calculated by dividing the number of achieved points by the number of items the video was evaluated on in each section. PEMAT-A/V scores range from 0% to 100%, with higher values generally indicating better understandability or actionability.

Accuracy, Utility, and Reliability

The accuracy, utility, and reliability of each video source were explored according to the JAMA (Journal of the American Medical Association) benchmark criteria [23]. These four criteria included authorship (ie, authors, contributors, affiliations, and credentials), attribution (ie, references and sources used for the content and copyright information), disclosures (ie, sponsorship, advertising, commercial funding, and potential conflicts of interest), and currency (ie, dates of posted and updated information). Each item can be scored as 0 ("disagree") or 1 ("agree"). Next, we calculated percentages of fulfilled items. The higher the value, the more accuracy, utility, and reliability elements were fulfilled.

Harms and Benefits

In order to summarize their potential benefit or harm, the videos were rated on an adapted 3-point scale as to whether they were perceived to be useful, neutral, or harmful for potential audiences [24]. Useful videos were judged to contain correct information and to be of value to patients, whereas harmful videos contained misleading or false information.

Statistical Analysis

Statistical analyses were conducted using SPSS Statistics for Windows (version 24; IBM Corp). Descriptive analyses included mean (SD) or median (range). Subgroup differences were explored using the Kruskal-Wallis test. The relationship between the individual items of the tests was examined using Spearman correlation. Statistical significance was set at $P \leq .05$. The interrater agreement of the two reviewers was determined using the intraclass correlation coefficient, as well as by determining the interitem correlation, r, between the individual reviewers.

Results

Video Identification and Baseline Characteristics

Our search identified 659 videos. Following a multistep process, three review authors (TS, MH, and LR) screened the videos for duplicates and checked them for compliance with the predefined eligibility criteria. Finally, 41 individual videos were considered for assessment (Figure 1). Most videos were provided by health professionals (15/41, 37%), followed by laypersons (6/41, 15%) and health portals (6/41, 15%). Furthermore, 10% of the videos (4/41) were offered by educational providers, and 7% (3/41) of the videos were TV reports uploaded by official TV channels or reuploaded by private providers. Out of 41 videos, 2 (5%) providers remained unclear.

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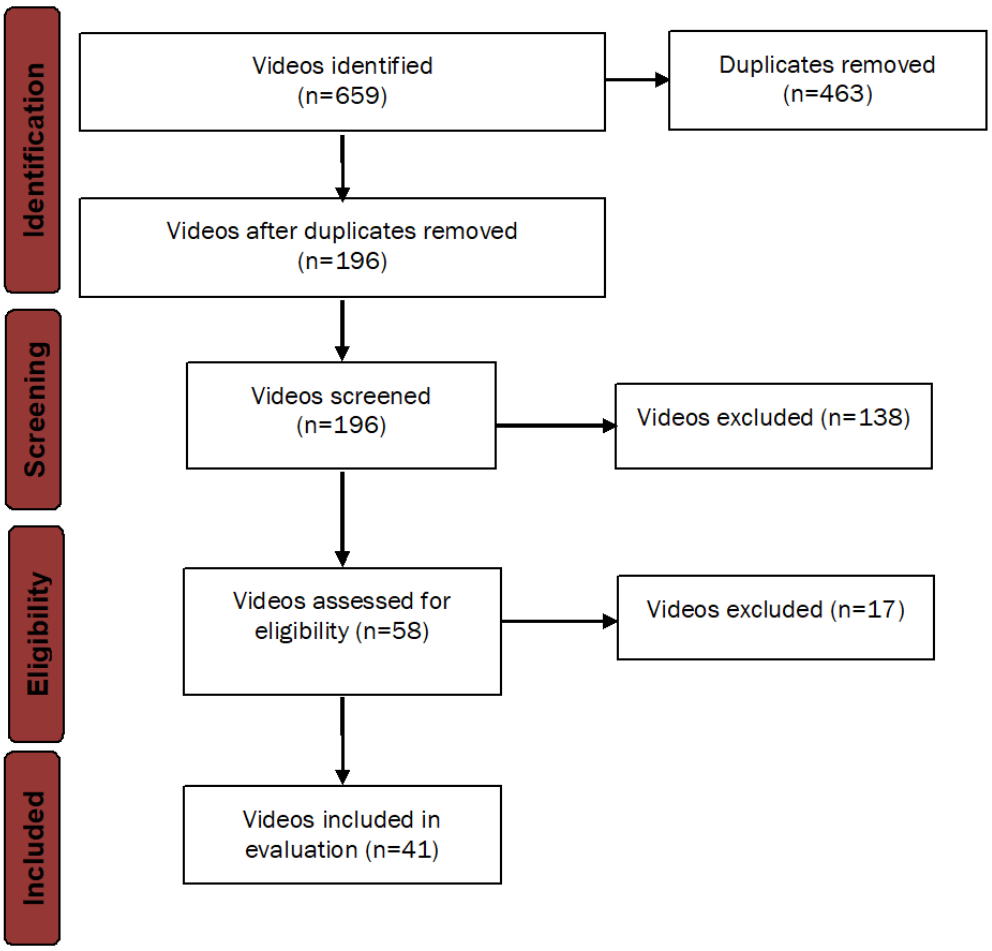
Figure 1. Flowchart showing the identification process of the videos.



The videos were uploaded between 2011 and 2020, with the majority (30/41, 73%) uploaded after 2017 (Table 1 and Multimedia Appendix 2). The number of views ranged from 25 to 386,195, with a mean of 27,853 views. The video length (minutes: seconds) ranged from 1:04 to 91:36. In 78% (32/41) of the videos, the duration was less than 10 minutes. The number of likes ranged from 0 to 17,925, with a median of 22. Most likes were given on a video dealing with the personal BCC history of a German influencer (video #20). The number of dislikes ranged from 0 to 333. The VPI was evaluable for 33 videos and ranged from 40 to 100.

Overall, video #8 ("Hautkrebs - Ein Überblick über Typen und Therapien"; Multimedia Appendix 2), provided by health professionals, and video #35 ("Weißer Hautkrebs – ein Patienteninformationsfilm"), created by a professional society, were rated best among all videos. Both videos gave an overview on the disease course. In contrast, video #30 ("Verjüngung mit Uta Baranovskyy: Weißer Hautkrebs Teil 3") provided by a layperson was rated the worst due to misleading information regarding the treatment of BCC.

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Table 1. Overview of baseline characteristics, quality, understandability, actionability, and reliability of the videos according to the respective categorization of the provider.

Characteristic	Provider								
	All	Layperson	TV reupload by private account	Health prof.[a]	Official TV report	Educ.[b] provider	Prof. society or NC[c] provider	Health portal	Unclear
Videos, n (%)	41 (100)	6 (15)	3 (7)	15 (37)	3 (7)	4 (10)	2 (5)	6 (15)	2 (4.9)
Views, mean (SD)	27,853 (70,693)	101,427 (1,591,378)	46,049 (77,209)	4221 (3105)	68,499 (57,295)	2177 (2751)	37,602 (25,232)	6980 (6836)	326 (350)
Video length (min:s), mean (range)	11:12 (1:04-91:36)	15:07 (1:15-37:44)	6:31 (4:20-9:12)	8:25 (1:16-65:50)	12:41 (4:04-29:13)	5:30 (1:04-16:52)	25:51 (8:42-43.01)	2:48 (1:49-3:57)	46:55 (2:15-91:36)
Year of upload, range	2011-2020	2017-2019	2011-2018	2012-2020	2012-2020	2016-2019	2016	2013-2020	2018-2020
Likes, mean (range)	672 (0-17,925)	4383 (20-17,925)	54 (0-139)	19 (0-59)	172 (53-344)	14 (0-42)	24.50 (0-49)	26 (0-68)	3 (2-3)
Dislikes, mean (range)	15 (0-333)	74 (0-333)	12 (0-34)	1 (0-3)	28 (12-51)	1 (0-3)	5 (0-9)	2 (0-5)	0 (0)
DISCERN score[d], mean (SD)	3.30 (0.80)	1.95 (0.41)	3.50 (0.44)	3.64 (0.53)	3.08 (0.94)	3.33 (0.41)	3.72 (0.93)	3.46 (0.70)	4.03 (0.04)
GQS score[e], mean (SD)	3.76 (1.13)	1.83 (0.41)	4.12 (0.29)	4.47 (0.64)	3.33 (1.26)	3.50 (0.41)	4.00 (0.71)	3.83 (1.21)	4.25 (1.06)
PEMAT-A/V score[f] (% U), mean (SD)	70.84 (13.32)	60.28 (11.50)	76.77 (10.16)	75.32 (13.26)	78.59 (11.33)	71.44 (11.49)	74.66 (18.16)	61.55 (10.92)	71.14 (19.61)
PEMAT-A/V score[f] (% A), mean (SD)	45.94 (43.74)	55.56 (50.18)	27.78 (25.46)	56.67 (40.21)	33.33 (33.33)	25.00 (50.00)	50.00 (70.71)	36.11 (37.14)	50.00 (70.71)
JAMA score[g] (%), mean (SD)	27.74 (22.10)	12.50 (15.81)	20.83 (7.22)	30.00 (16.23)	37.50 (21.65)	43.75 (38.86)	62.50 (17.67)	12.50 (15.81)	31.25 (26.52)

[a]prof: professional.

[b]educ: educational.

[c]NC: noncommercial.

[d]DISCERN items were scored on a 5-point scale ranging from 1 (“criterion is not met at all”) to 5 (“criterion is fully met”); videos were considered good quality (≥4), medium quality (≥2 to <4), or low quality (<2).

[e]GQS: Global Quality Scale; the GSQ was scored a 5-point scale ranging from 1 (“low quality”) to 5 (“high quality”); videos were considered high quality (4 or 5), medium quality (3), or low quality (1 or 2).

[f]PEMAT-A/V: Patient Education Materials Assessment Tool for Audiovisual Materials; scores range from 0% to 100%, with higher values indicating better understandability (U) or actionability (A).

[g]JAMA: Journal of the American Medical Association; each of four criteria were scored as 0 (“disagree”) or 1 (“agree”); scores range from 0% to 100%, with higher values indicating higher reliability.

Quality: DISCERN and GQS Results

Out of 45 points in total, the 41 individual videos ranged between 10.5 and 35.0 points according to the DISCERN tool. The mean DISCERN scores per video ranged from 1.31 to 4.38 points, with an average mean score of 3.31 (SD 0.80) points, indicating medium quality (Table 1). Most score deductions were due to lacking information about the sources used to create the respective video or missing complementary information. The mean GQS score was 3.8 (SD 1.1) points, indicating medium quality as well.

Understandability and Actionability: PEMAT-A/V Results

The average PEMAT-A/V score was 70.84% (SD 13.32%, range 43.18%-100%) for understandability and 45.94% (SD 43.74%, range 0%-100%) for actionability. Most score deductions for the understandability domain were due to a lack of a summary and because no visual aids were deployed. For the actionability domain, information was often missing regarding the interpretation of certain figures in order to take action.

Accuracy, Utility, and Reliability: JAMA Results

In total, a mean of 27.74% (SD 22.1%, range 0%-87.5%) of the JAMA benchmark criteria were fulfilled, indicating rather poor

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reliability. The main reasons for score deductions were missing information regarding the currency of videos (ie, the upload date) and missing disclosure of the provider.

Harms and Benefits

A total of 49% (20/41) of the videos were evaluated as useful, 7% (3/41) were evaluated as harmful, and the remaining videos were evaluated as neither beneficial nor harmful. All videos estimated to be harmful were provided by laypersons.

Interrater Agreement

We calculated intraclass correlation coefficients ranging from 0.940 to 0.955 with a Cronbach α of .973, indicating high overall interrater agreement concerning the assessments by the DISCERN tool, the GQS, the JAMA criteria, and the PEMAT-A/V. The interitem correlation, *r*, was 0.949, indicating high individual agreement among the two reviewers when assessing the individual items.

Subgroup Analyses

Significant differences in video quality, according to the DISCERN tool and the GQS, were identified between videos provided by laypersons and health professionals (P=.01; ie, videos by health professionals were judged as having higher quality than those provided by laypersons).

Regarding the assessment of whether videos were beneficial or not, differences were found in terms of the quality of the videos. Videos rated as beneficial showed significantly better quality in comparison to those rated as harmful (DISCERN: P=.004; GQS: P=.002) and neutral (DISCERN: P=.006; GQS: P<.001), according to the DISCERN tool and the GQS. No further subgroup differences were identified.

Correlation Analysis

A significant positive correlation was found between DISCERN and GQS values (r=0.836) as well as between DISCERN values and reliability and understandability criteria (r=0.488 and r=0.460, respectively; Table 2). In addition, the quality according to the GQS also significantly correlated with the reliability (r=0.426) and understandability (r=0.482) of the videos. Furthermore, the longer the duration of a video, the more understandability (r=0.454) and actionability (r=0.314) items had been deployed. No further significant correlations between the baseline characteristics and the quality, reliability, understandability, or actionability of the videos were identified.

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Table 2. Correlation analysis (Spearman *r* and two-tailed *P* value) among the research variables.

Variable	Baseline characteristics					Quality		Reliability	PEMAT-A/V[a]	
	Likes	Dislikes	Views	Duration	Uploads	DISCERN	GQS[b]	JAMA[c]	Understandability	Actionability
Baseline characteristics										
Likes										
r	1	0.784[d]	0.604[d]	0.375[e]	–0.060	–0.085	–0.082	–0.084	0.031	0.215
P value	__[f]	<.001	<.001	.02	.71	.60	.61	.60	.85	.18
Dislikes										
r	0.784[d]	1	0.619[d]	0.228	–0.323[e]	–0.186	–0.151	0.037	–0.119	0.066
P value	<.001	—	<.001	.15	.04	.25	.35	.82	.46	.68
Views										
r	0.604[d]	0.619[d]	1	0.200	–0.380[e]	0.086	0.003	0.068	0.029	0.055
P value	<.001	<.001	—	.21	.01	.59	.99	.67	.86	.74
Duration										
r	0.375[e]	0.228	0.200	1	0.226	–0.013	–0.025	0.115	0.454[d]	0.314
P value	.02	.15	.21	—	.16	.93	.87	.47	<.001	.06
Uploads										
r	–0.060	–0.323[e]	–0.380[e]	0.226	1	0.059	0.106	–0.152	0.300	0.157
P value	.71	.04	.01	.16	—	.72	.51	.34	.06	.33
Quality measures										
DISCERN										
r	–0.085	–0.186	0.086	–0.013	0.059	1	0.836[d]	0.488[d]	0.460[d]	0.135
P value	.60	.25	.59	.93	.72	—	<.001	<.001	<.001	.40
GQS										
r	–0.082	–0.151	0.003	–0.025	0.106	0.836[d]	1	0.426[d]	0.482[d]	0.186
P value	.61	.35	.99	.87	.51	<.001	—	<.001	<.001	.24
Reliability measures										
JAMA										
r	–0.084	0.037	0.068	0.115	–0.152	0.488[d]	0.426[d]	1	0.469[d]	–0.052
P value	.60	.82	.67	.47	.34	<.001	<.001	—	<.001	.75
PEMAT-A/V measures										
Understandability										
r	0.031	–0.119	0.029	0.454[d]	0.300	0.460[d]	0.482[d]	0.469[d]	1	0.220
P value	.85	.46	.86	<.001	.06	<.001	<.001	<.001	—	.17
Actionability										
r	0.215	0.066	0.055	0.314[e]	0.157	0.135	0.186	–0.052	0.220	1
P value	.18	.68	.74	.05	.33	.40	.24	.75	.17	—

[a]PEMAT-A/V: Patient Education Materials Assessment Tool for Audiovisual Materials.

[b]GQS: Global Quality Scale.

[c]JAMA: Journal of the American Medical Association.

[d]The correlation is significant at a significance level of <.001 (two-tailed).

[e]The correlation is significant at a significance level of .05 (two-tailed).

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[f]Not applicable.

Discussion

Principal Findings

In this study, 41 YouTube videos about BCC have been systematically identified and evaluated by two independent reviewers. For the first time, we present an in-depth and objective assessment of the quality, understandability, and reliability of the information about BCC provided by YouTube videos on this subject. There were more than 1 million views among the 41 videos identified in our search, highlighting the importance of the internet and platforms like YouTube as sources of health information. Half of the assessed videos were estimated to be beneficial for patients, showing that YouTube may be an important tool for information broadcasting. The percentage of beneficial videos was similar compared to the results of previous studies evaluating video contents about other diseases [25-27].

Our results complement the currently available evidence on informational material available for other types of skin cancer, such as videos, brochures, or websites [14,27,28]. Our evaluation shows that currently available BCC videos were, overall, of medium to good quality and understandability but had low actionability and poor reliability. In addition, we have shown that videos of longer duration applied more understandability and actionability items and that the quality of videos provided by health professionals was significantly higher than that of videos provided by laypersons.

Interestingly, none of the videos identified in our search were provided by pharmaceutical companies, which sharply contrasts with our previous search and evaluation of videos on melanoma [27]. In that study, 16% of the videos had been created by pharmaceutical companies and nearly one-third by laypersons, while most videos on BCC had been supplied by health professionals. A potential explanation might be that pharmaceutical companies offer more videos on melanoma, as the interest in disease-specific knowledge is judged to be more important due to the complexity and abundance of different therapy regimens. Nevertheless, our evaluation revealed that videos about BCC provided by health professionals scored the best ratings in terms of quality, understandability, and reliability. This may be explained by the fact that these providers have better resources and scientific backgrounds to produce such high-quality videos.

In summary, the quality, understandability, and reliability of the BCC videos were comparable to those about melanoma [27]. However, BCC videos were judged to score more points on actionability items and fewer points on reliability items. Notably, the most likes were awarded for the two videos uploaded by a female influencer describing her own personal history with BCC as well as her therapy and follow-up. While these videos were mostly inferior in comparison to other videos, they highlight that the involvement of testimonials or influencers might be a feasible approach to maximize the awareness of skin cancer, in general, and to promote preventive measures. However, on the other hand, they may also use their coverage to distribute incorrect or harmful information.

YouTube is a growing online video platform providing easy access [12] with steadily increasing popularity among patients and medical professionals [29]. Distribution of medical information to such a huge audience offers invaluable opportunities but also risks of misinformation and biased presentation. Since the accuracy of online information is variable and since there is no peer review of such videos, the credibility and trustworthiness of the providers cannot be verified [15-17]. Moreover, quality certificates, like HONcode (Health on the Net Foundation Code of Conduct), which are awarded for reliable health-related webpages, are missing for YouTube videos [30]. Additionally, YouTube can be used as an advertising tool. As users can share their personal opinions without sufficient information and experience, videos may mislead patients and affect the physician-patient relationship [31]. Obtaining correct information from reliable sources is crucial, as it increases patients' satisfaction and empowerment and may improve treatment results [32,33]. Efforts should be undertaken to introduce regular quality control of videos with medical content on YouTube.

We are aware that this study has some limitations. YouTube search results are highly dynamic and will change when new videos are uploaded and when old videos are removed. Additionally, we did not include videos with restricted access (eg, asking for log-in information).

Conclusions

Overall, our study demonstrates that online videos on BCC are currently of medium to good quality and are predominantly uploaded by health professionals. However, the reliability of the videos was poor. As more and more patients use online material, including YouTube videos, for acquiring disease-specific knowledge, it is crucial to ensure good quality, understandability, and reliability prior to publication. Thus, optimization of the videos is desirable. In particular, adaptation to reliability criteria is necessary to support patient education and increase transparency. Patients should be advised to check the sources of the videos and whether their content is up to date.

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Conflicts of Interest

None declared.

Multimedia Appendix 1
Overview of the different assessment tools used for the evaluation of the videos.
[DOCX File , 18 KB - cancer_v8i1e29581_app1.docx]

Multimedia Appendix 2
Summary of information about 41 videos on melanoma in German analyzed in this study.
[DOCX File , 31 KB - cancer_v8i1e29581_app2.docx]

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Abbreviations

BCC: basal cell carcinoma
GQS: Global Quality Scale
HONcode: Health on the Net Foundation Code of Conduct
JAMA: Journal of the American Medical Association
N/A: not applicable
PEMAT-A/V: Patient Education Materials Assessment Tool for Audiovisual Materials
VPI: video power index

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Original Paper

Automated Clinical Practice Guideline Recommendations for Hereditary Cancer Risk Using Chatbots and Ontologies: System Description

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Abstract

Background: Identifying patients at risk of hereditary cancer based on their family health history is a highly nuanced task. Frequently, patients at risk are not referred for genetic counseling as providers lack the time and training to collect and assess their family health history. Consequently, patients at risk do not receive genetic counseling and testing that they need to determine the preventive steps they should take to mitigate their risk.

Objective: This study aims to automate clinical practice guideline recommendations for hereditary cancer risk based on patient family health history.

Methods: We combined chatbots, web application programming interfaces, clinical practice guidelines, and ontologies into a web service–oriented system that can automate family health history collection and assessment. We used Owlready2 and Protégé to develop a lightweight, patient-centric clinical practice guideline domain ontology using hereditary cancer criteria from the American College of Medical Genetics and Genomics and the National Cancer Comprehensive Network.

Results: The domain ontology has 758 classes, 20 object properties, 23 datatype properties, and 42 individuals and encompasses 44 cancers, 144 genes, and 113 clinical practice guideline criteria. So far, it has been used to assess >5000 family health history cases. We created 192 test cases to ensure concordance with clinical practice guidelines. The average test case completes in 4.5 (SD 1.9) seconds, the longest in 19.6 seconds, and the shortest in 2.9 seconds.

Conclusions: Web service–enabled, chatbot-oriented family health history collection and ontology-driven clinical practice guideline criteria risk assessment is a simple and effective method for automating hereditary cancer risk screening.

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KEYWORDS

service-oriented architecture; restful API; hereditary cancer; risk assessment; clinical practice guidelines; consumer health informatics

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Introduction

Identifying Patients at Risk of Hereditary Cancer is Challenging

Family health history (FHx) is the most important indicator of the risk of hereditary cancer [1-3]. However, providers have insufficient time to collect and analyze FHx during a patient visit and lack confidence and training in assessing FHx for hereditary cancer risk [4-6]. In addition, the clinical practice guidelines (CPGs) used to assess patient FHx for hereditary cancer risk are numerous and complicated. Many patients with FHx indicative of hereditary cancer risk are unreliably and inaccurately referred for cancer genetic consultation services or are missed altogether [7-9]. Even with accurate FHx collection and assessment, there is a shortage of genetic counselors to meet the needs of cancer genetic consultation services [10,11]. Patients and providers need help in collecting and assessing FHx for hereditary cancer risk to identify patients at risk for earlier counseling and preventive efforts.

CPGs and FHx Are Important Tools for Identifying Patients at Risk

CPGs contain criteria that amalgamate and organize clinical knowledge relevant to hereditary cancer syndromes. They also define thresholds, curated by experts, based on clinical knowledge for making referral recommendations for cancer genetic counseling and testing [12-15]. There are several organizations that publish these guidelines for various cancer syndromes with varying frequencies, including but not limited to the National Comprehensive Cancer Network (NCCN), the American College of Medical Genetics and Genomics (ACMG), and the US Preventive Services Task Force. Although CPGs are curated by panels of experts and do not necessarily constitute validated tools, they are valuable reference points for considering hereditary cancer risk based on FHx and, if efficiently applied across the patient population, could serve as a valuable indicator of potential risk.

The most useful tool for evaluating whether FHx meets CPG criteria is the family pedigree—a chart with connected squares (males) and circles (females) that depicts family members, their relationships, cancer diagnoses, age, and other relevant information [16]. Risk conveyed to the proband depends on the relationship the proband has with affected and unaffected relatives in their pedigree. The knowledge required to assess CPG criteria can be represented using an ontology. An ontology is a formal, explicit specification of a shared conceptualization [17]. In other words, an ontology is a machine-readable representation of shared knowledge on which thresholds can be evaluated to determine whether FHx meets the criteria for any given patient. Efficiently intersecting family pedigrees and CPG criteria is a necessary step in making timely referral recommendations for cancer genetic consultations.

Previous Decision Support Tools for Applying CPGs to Patient Data

Over the past several decades, various projects and tools have been built to model and encode CPGs in an effort to increase their value within the clinical workflow. Athena DSS developed at Stanford [18], Asbru developed at Stanford [19], GEM developed at Yale [20,21], GLIF3 [22,23], EON developed at Stanford [24], PROforma developed by John Fox at the Imperial Cancer Research Fund [25], GUIDE [26], Prodigy [27], and, more recently, Sharable Active Guideline Environment (SAGE) [28,29], are all technologies that have been devised to computerize CPGs for hypertension, diabetes, immunization, and others. These tools range widely from XML-based document models to clinical workflow–driven decision support systems designed to formalize CPG knowledge, manage temporal constraints, and integrate with clinical workflows and systems. SAGE, the most recent of these systems, used Protégé [30], an ontology development tool developed at Stanford University, to represent CPG knowledge in ontologies [29].

Ontologies are useful tools for modeling and representing knowledge. More than simple databases, ontologies define concepts and relationships about which inferences can be made beyond logical or statistical measures of the data. Well-known biomedical ontologies that support medical billing, coding, and research include Systematized Nomenclature of Medicine–Clinical Terms [31,32] and the National Cancer Institute Thesaurus [33]. Perhaps the biggest challenge in using ontologies to provide value to clinical care is their size and the processing power required to apply them to patient data. Domain ontologies, as proposed by Musen [34], are designed to overcome this challenge by scoping the ontology to a specific set of concepts and relationships within an application area, such as hereditary cancer. He argues that separating medical knowledge into domain ontologies empowers domain-independent problem solvers, such as software applications in medical informatics, to solve application-level tasks, such as applying CPGs to FHx and recommending genetic counseling.

Unlike the CPG modeling technologies and the ontologies described above, which are designed to be used within clinical workflows and integrate directly with electronic health record (EHR) systems, collecting FHx and assessing hereditary cancer risk are largely agnostic of clinical workflows. Furthermore, FHx in EHR systems is notoriously poor [35,36]. FHx is collectively held by the patient and their family members, which adds to the complication of maintaining accurate FHx within an EHR [37]. Applying CPGs for conditions or domains that depend primarily on data points specific to the patient requires a system that can make the right recommendation at the right time in the clinical workflow based on changing values in a patient's EHR. However, FHx does not really change a great deal from visit to visit and depends heavily on information the patient may not have during a patient-provider consultation. In addition, as previously pointed out, there is not sufficient time to collect and assess FHx during a patient-provider visit. These challenges necessitate a solution that emphasizes patient involvement and ownership of their family history collection and assessment before visiting their health care provider.

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Web Application Programming Interfaces and Chatbots Increase Patients' Access to CPG Recommendations

Chatbot-oriented FHx collection and ontology-driven CPG risk assessment implemented in a web service architecture have the potential to empower patients through a simple and effective mechanism for automating initial FHx collection and risk assessment. In previous studies, we demonstrated the utility of collecting FHx using chatbots and web services, the most recent of which engaged >10,000 individuals in collecting and assessing their FHx [38,39]. Research has shown that although conversational chatbot agents can take a little longer to interact with, users reported higher overall satisfaction, perceived usefulness of the system, perceived quality of information collected, and significantly better interface quality, with 3 out of 4 users preferring chatbots to traditional data collection methods [40,41]. The chatbot we built for the system is a workflow-driven chatbot that follows a branching logic strategy for optimal user experience [39,42]. The observed participation in collecting FHx using chatbots is evidence that patients with cancer in their families are motivated to learn about their risk. Once patients collect their FHx using the chatbot, they only need access to CPG criteria for initial risk assessment that does not require the time and attention of a trained professional.

Access is best provided to formalize CPG knowledge for risk assessment using web application programming interfaces (APIs) that can receive electronic FHx data and return CPG recommendations. Web APIs, or representational state transfer APIs, form the underpinnings of modern web development by providing access to data, processes, and information on the web in a general, scalable, and secure manner through the browser [43]. By combining chatbots and ontological representations of CPGs for FHx with web APIs, patients can collect their FHx and receive CPG recommendations from the comfort of their own home with their family members and share their results with their provider at a future provider consultation.

The objectives of our study are to collect and store FHx in an electronic format, organize CPGs into a knowledge representation that can be applied to the FHx, and design a system that can assess FHx using CPG criteria and return the relevant recommendations using web APIs. This paper describes the ontological representation of hereditary cancer CPGs from NCCN and ACMG and the system that applies the CPG ontology to patient FHx to determine whether cancer genetic consultation should be considered. This paper will help biomedical informaticists and web application developers understand how to automate the application of domain ontologies to patient data using ontology programming interfaces (OPIs) and web APIs.

Methods

Hereditary Cancer CPG Ontology

Overview

The hereditary cancer CPG ontology was developed by JBR and reviewed by LF. We selected criteria from the ACMG and NCCN hereditary cancer CPGs for the most prevalent cancer syndromes. These criteria outlined the domain knowledge necessary to create an ontological representation of CPGs and write rules in the ontology that represent the CPG criteria. Ontologies were developed using Python (version 3.7), the Protégé ontology editor (version 5.2.0), and the Owlready2 OPI (version 2.21), which includes a modified version of the HermiT Reasoner developed by the Department of Computer Science at the University of Oxford [44]. The ontology was designed to represent all possible states for patient FHx according to the CPGs as efficiently as possible. We used Owlready2 to dynamically generate and modify the ontology in Python using JSON data structures. The system is open source and available on Bitbucket [45].

Representing CPGs Using Ontologies

Hereditary cancer domain knowledge from the CPGs is defined in the ontology using concepts, properties, and individuals. Relationships between concepts are defined in the ontology by using Resource Description Framework triples and equivalency classes to represent CPG criteria that can be applied to FHx instantiated as individuals in the ontology. For example, consider a family history where the proband has a father and a brother both diagnosed with prostate cancer before the age of 55 years. When the patient engages the chatbot, the family member workflow will ask which family members had cancer and what age they were diagnosed (Figure 1). On the basis of the answers to these questions, which directly tie to the ontology logic described below, this proband is considered at risk and should consider a cancer genetic consultation based on the following criterion in ACMG: ≥2 cases of prostate cancer diagnosed at age ≤55 years in close relatives. This criterion is modeled in the ontology in two separate subclasses of ACMGProstatePatient:

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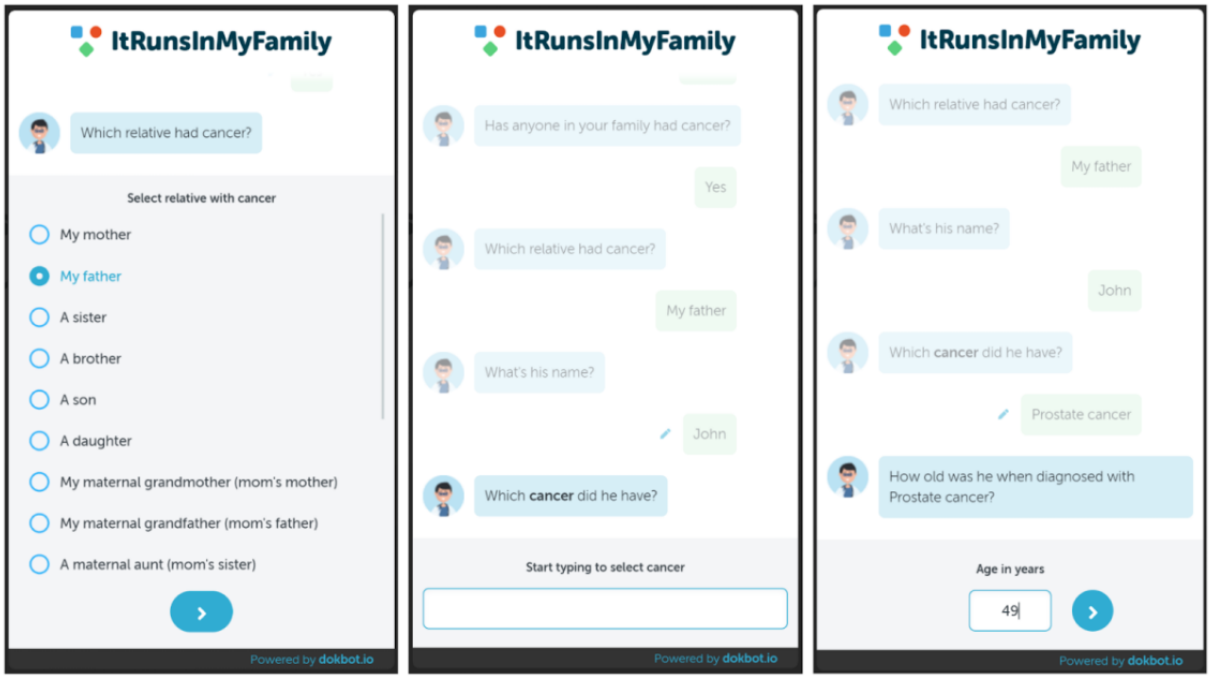
Figure 1. The chatbot collects family health history (FHx) relevant to CPG ontology logic. In this example, the patient enters values for an FHx where the father has prostate cancer before the age of 55 years. The workflow will also collect the same data for all other family members. Per the example in the text, if both father and a brother of the proband have prostate cancer before the age of 55 years, they would meet the ACMG criterion ≥2 cases of prostate cancer diagnosed at age ≤55 years in close relatives. ACMG: American College of Medical Genetics and Genomics; CPG: clinical practice guidelines.



Prostate.01 accounts for the cases where the proband and a close family member have prostate cancer diagnosed before the age of 55 years. Prostate.02 accounts for cases where the proband does not have cancer but 2 close family members have prostate cancer diagnosed before the age of 55 years. An important rule that applies to all criteria is that rules involving ≥1 family member must be on the same side of the family to truly evaluate hereditary patterns. The aforementioned example rules account for this by considering that the relationships *has_maternal_close_relative* and *has_paternal_close_relative.* Together, the aforementioned rules represent the prostate criterion that our example proband meets. When the reasoner classifies the proband as a subclass of the ACMGProstatePatient, the system knows to recommend a genetic cancer consultation for the proband. Equivalencies such as Prostate.01-02 capture CPG criteria and are the crux of automated identification of patients at risk of hereditary cancer. A list of all criteria implemented from ACMG and NCCN can be found in Tables S1 and S2 in the Multimedia Appendix 1.

Ontology Construction

To successfully apply the CPG ontology to patient data, we recognized that certain design patterns were necessary to ensure reasonable processing time and out-of-the-box application of the HermiT Reasoner. First, only CPG knowledge concepts necessary for applying the CPG criteria should be included in the CPG domain ontology to prevent bloat and ensure acceptable reasoning times with the HermiT Reasoner. Therefore, concepts related to treatment, for example, are not included. Second, the CPG criteria rules should be contained as subclasses of the *Patient* class, thereby ensuring that after a patient's FHx is instantiated in the ontology and the reasoner has completed reasoning, the patient has been reclassified within the ontology under *Patient* subclasses that correspond to the CPG criteria met by their FHx, for example, ACMGProstatePatient. The result is a lightweight, patient-centric domain ontology that is readily adapted to run inside a web API.

Ontology construction is an iterative process that relies heavily on the CPGs to determine concepts, relationships, and equivalencies to be defined in the ontology. Throughout the development process, the criteria interpretations in the ontology equivalency classes were reviewed by a genetic counselor (CB) and an oncologist (JDS). As new classes were added to the ontology, test cases were created to ensure that the equivalency classes worked as expected.

FHx assessment depends on how many family members on one side of the family are diagnosed with certain combinations of cancers at or before specific ages in the presence of specific disease factors. The thresholds defined by the CPGs are minimum thresholds that require frequent use of the cardinality restriction *MIN*. Ontological reasoning with cardinality restrictions is complex and time consuming. Other methods, such as SPARQL Protocol and RDF Query Language (SPARQL) queries, do not handle cardinality restrictions easily. Handling cardinality restrictions with SPARQL is difficult as if one considers a cardinality restriction with a cardinality of n, one needs to (1) search the n relations, (2) verify that they are all distinct, and (3) remove duplicates (eg, if n=2 and one finds the a,b relation, then b,a should not be considered as a distinct result). This typically requires many triples in SPARQL, especially if the value of the cardinality restriction is complex (for example, another restriction), as in that case, it must be copied n times in the SPARQL query. Another option is to use a *GROUP BY* statement in SPARQL. However, this allows only a single cardinality restriction. If there is ≥1 such restriction, it

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would require us to run multiple separate queries and then take the union of their results [46]. Thus, we used an ontological definition as we had to rely heavily on the MIN cardinality restriction to implement the criteria. To keep reasoning times down, we frequently used ≥1 equivalency class to represent a single CPG criterion.

Owlready2 and Ontology-Oriented Programming

Owlready2 is a lightweight Python library designed to programmatically create and edit ontologies [47]. By using Python's inherent hierarchical class structure, Owlready2 provides an intuitive OPI for ontology-oriented programming. In addition, Owlready2 is able to bind specific programmatic functionality to ontology concepts by declaring Python functions directly within ontology classes. This ability makes it possible to treat ontology classes such as objects in object-oriented programs. Web APIs are generally built using object-oriented programming development patterns and often control interactions with relational databases through an object relational mapper (ORM) [48,49]. ORMs are the link between web APIs and relational databases that provide access and management of data in the database via the web API. We used Owlready2 as a key resource in developing the CPG ontology but, more importantly, as a kind of ORM for interfacing with the ontology to provide our web API access to the knowledge in the ontology for FHx cases.

System Evaluation and Testing

We created 192 test cases—at least one test per CPG criterion—to evaluate the system and ensure equivalency classes performed as expected. Each test case had a target and a payload. The target is the correct recommendation in the CPG ontology, and the payload is the test case FHx. The FHx in the payload adheres to the same JSON schema as the FHx received by the chatbot (Figure 2). The creation of test cases was heavily driven by the criteria for which the test was written. Each test case was built to reflect combinations of family cancer diagnoses, disease factors, and ages of onset to trigger the target CPG criterion.

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Figure 2. Example JSON family health history (FHx) format, JSON is a ubiquitous data structure for web development based on key value pairs. Each object in the relatives list represents a family member with the exact same FHx format as the proband. This example only shows factors for breast cancer.

```
{
    "fullName": "str",
    "relationToProband": "str",
    "age": int,
    "sex": "str",
    "conditions": [
        {
            "conceptId": "str",
            "ageOnset": int,
            "factors": {
                "laterality": "str",
                "cancerTissueOrigin": [],
                "erStatus": "str",
                "prStatus": "str",
                "her2Status": "str"
            }
        }
    ],
    "relatives": [
        {
            "fullName": "str",
            "relationToProband": "str",
            "age": int,
            "sex": "str",
            "conditions": [],
            “traits”: {}
        }
    ],
    "traits": {
        "ancestry": []
    }
}
```

System Architecture

Overview

Patient FHx collected by the chatbot [39] is sent to a web API (FHx API) that manages access to the CPG ontology. The FHx API is responsible for instantiating the FHx using the CPG ontology, initiating the HermiT Reasoner to apply CPG criteria, and retrieving final recommendations. The HermiT Reasoner is a state-of-the-art ontology reasoner that is packaged with most ontology development resources such as Protégé and Owlready2 [50]. The FHx API then sends the results to another web API (report API) that packages the information into a PDF report. The report API is capable of sending the report to the patient or to the patient’s provider.

FHx API Components

The FHx API has an ontology access object (OAO) layer, a service layer, and a reasoning layer (Figure 3). The OAOs coordinate access to the ontology for all other API components. The ontology service is the most important part of the FHx API and is responsible for providing access to the CPG ontology, all OAOs, and the HermiT Reasoner. There are two other main services: Patient service and Cancer service. These 2 services are specifically named after the Patient and Cancer classes in the CPG ontology and have corresponding OAOs. Importantly, they each have access to the ontology service to coordinate with their respective OAOs to instantiate FHx using the ontology and initiate reasoning. In addition, they use their OAOs to retrieve recommendation results after the HermiT Reasoner completes.

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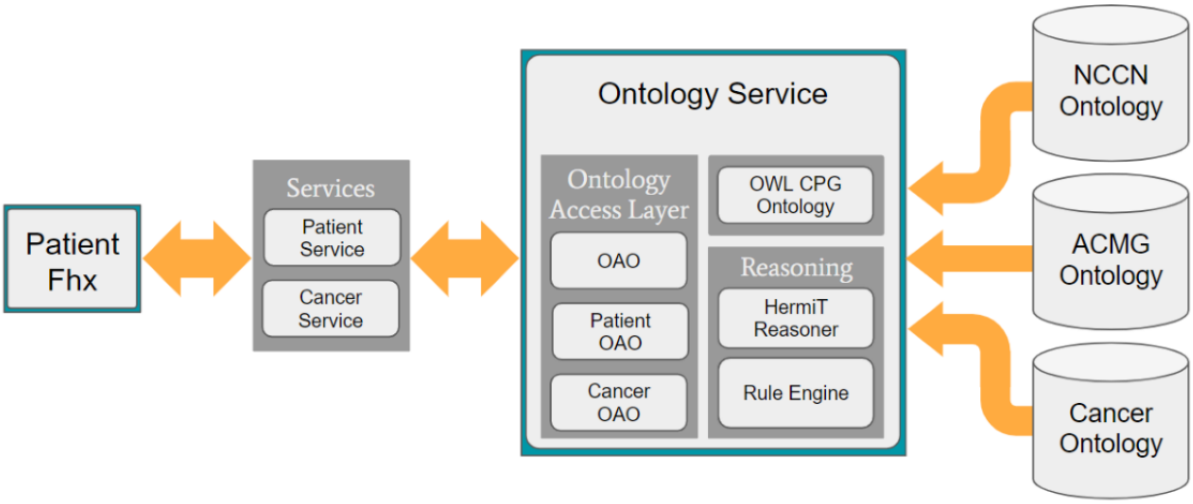
Figure 3. System architecture. Patient family health history (FHx) is received by the system; services use the ontology service to model patient data using the CPG ontology, perform reasoning, and make recommendations. ACMG: American College of Medical Genetics and Genomics; CPG: Clinical practice guidelines; NCCN: National Comprehensive Cancer Network; OAO: ontology access object.



Instantiating Patient Data With Services and OAOs

Owlready2 allows Python functions to be declared within ontology classes, enabling object-oriented programming methods to be used to instantiate FHx using the CPG ontology. The OAO layer makes the most use of this by defining the methods for setting relationships and other important properties necessary for instantiating the FHx in the ontology. Each service in the service layer has an associated OAO wherein all Python functions that immediately access the ontology reside. For example, the Patient service receives patient FHx in JSON format and relies on the Patient OAO to add family members to the ontology as individuals, set family relations, and retrieve recommendations. An example patient FHx JSON format can be viewed in Figure 2. Separating service logic from OAOs isolates interactions with the ontology and emulates a well-established pattern of developing traditional web APIs where access to relational databases is encapsulated within database access objects. Once the patient FHx is instantiated using the CPG ontology, it is ready for the HermiT Reasoner to apply CPG criteria.

Reasoning and Retrieving Results With OAOs

After the patient's FHx has been instantiated by creating individuals in the ontology to represent the family members and their respective conditions, the ontology service calls the reasoning layer. Owlready2 uses the HermiT Reasoner to execute previously defined equivalency classes within the ontology that contains CPG criteria. Patient FHx instantiated within the ontology is reclassified accordingly to indicate which CPG criteria they meet, if any. Once the reasoning is completed, the service layer accesses the reclassified FHx from the ontology using OAOs and returns CPG-based recommendations (Figure 3). Importantly, the CPG ontology is reloaded for each FHx it evaluates to ensure that FHx from previous probands has been removed.

Results

Hereditary Cancer CPG Ontology

Overview

Using Python, Owlready2, and Protégé, we generated a hereditary cancer ontology with 758 classes, 20 object properties, 23 datatype properties, and 42 individuals and visualized it using WebVOWL [51] to produce a graph with 781 nodes and 1015 edges (Figure 4). The blue circles represent classes in the ontology class hierarchy, the blue boxes on lines between concepts represent object properties, and the green boxes on lines between the yellow boxes represent data properties and data types, respectively. The parent classes in the ontology class hierarchy are *Ancestry*, *Cancer*, *CancerGene*, *CancerTissueOrigin*, *DiseaseFactor*, *Histology*, *HormoneStatus*, *Laterality*, *Patient*, *Polyp*, *Sex*, *Syndrome*, and *Trait* and include 44 cancers, not including subtypes, 144 genes, 73 criteria from ACMG, and 40 criteria from NCCN. Static individuals in the ontology are represented by increasing the area of the concept they belong to in the graph. *Ancestry* (10 static individuals) is the largest, followed by *CancerTissueOrigin* (8 static individuals), *Histology* (6 static individuals), *HormoneStatus* (6 static individuals), *Trait* (4 static individuals), *Sex* (3 static individuals), *Laterality* (3 static individuals), and *DiseaseFactor* (2 static individuals).

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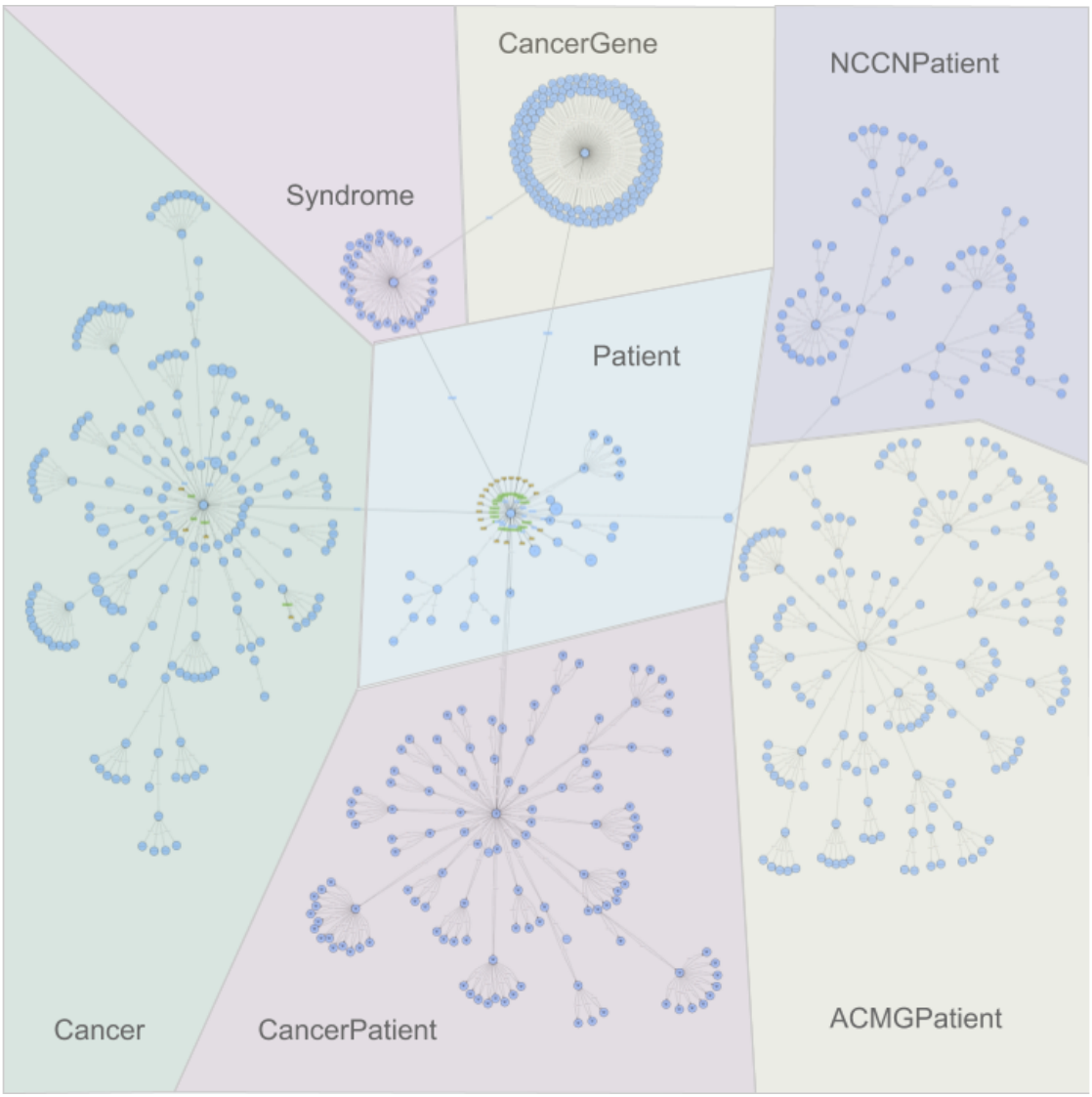
Figure 4. Ontology graph produced using WebVOWL. The Patient class is the central feature of the ontology and is linked to the *Cancer*, *Syndrome*, and *CancerGene* classes by the properties *has_cancer*, *has_syndrome*, and *has_mutation_in*, respectively. *Ancestry*, *CancerTissueOrigin*, *DiseaseFactor*, *Histology*, *HormoneStatus*, *Laterality*, *Polyp*, *Sex*, and *Trait* are concentrated around the Patient class. The right side of the graph represents the NCCNPatient class and the ACMGPatient class and their respective subclasses. ACMG: American College of Medical Genetics and Genomics; NCCN: National Comprehensive Cancer Network.



Patient Class

The most important class in the ontology is that of the *Patient* and every family member, including the proband, and is instantiated as an individual of the Patient class when a patient's FHx is processed. The most important subclasses of the Patient class are *CancerPatient* and *PatientWithRecommendations*. *CancerPatient* is used to define the proband and their family members in terms of the cancers they have, and *PatientWithRecommendations* is the parent class of *ACMGPatient* and *NCCNPatient classes*. All CPG criteria are housed in the equivalency subclasses of *ACMGPatient* and *NCCNPatient* and rely on the equivalency classes in *CancerPatient* and *Cancer* to evaluate the CPG criteria. The CPG criteria implemented from ACMG and NCCN can be found in Tables S1 and S2 in the Multimedia Appendix 1. In Figure 4, *Ancestry*, *Polyp*, *Sex*, and *Trait* are also inside the *Patient* block. Although they are not strictly subclasses of the *Patient* class, they represent small clusters of the ontology that are directly related to the *Patient* class.

The *PatientWithRecommendations* class is the parent class of all the guidelines implemented by the ontology. *ACMGPatient* and *NCCNPatient* represent 2 isolated clusters in the ontology that encapsulate separate but very similar hereditary cancer guidelines. Each leaf node in *ACMGPatient* and *NCCNPatient* represents a CPG criterion used to evaluate patient FHx. Breast, ovarian, pancreatic, colorectal, and endometrial cancer guidelines are implemented for both ACMG and NCCN, along with guidelines specific to Li–Fraumeni syndrome (LFS) and Lynch syndrome (LS). In addition, brain, gastric, melanoma, prostate, renal, and thyroid cancer guidelines are implemented for ACMG. This accounts for the relative size difference between the *ACMGPatient* and *NCCNPatient clusters*.

Cancer

Cancer is the next most important class in addition to the *Patient* class in the ontology class hierarchy for evaluating patient FHx.

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A total of 44 cancers (83 including subtypes) are represented in the *cancer* block in Figure 4. Although not all of them are immediately pertinent to hereditary cancer CPGs, it is important to include them in an accurate patient FHx. The most frequently used cancers by the CPGs are breast, ovarian, colorectal, endometrial, and those related to LFS and LS. In Figure 4, *CancerTissueOrigin, DiseaseFactor, Histology, HormoneStatus,* and *Laterality* are also inside the *cancer* block and are represented by static individuals. *CancerTissueOrigin* is specific to where the cancer originated in a patient, for example, *ductal* and *lobular* for breast cancer; *DiseaseFactor* includes factors for different cancers, for example, *mmr_stable; Histology* represents different histologies, most notably for kidney cancer, for example, *clear_cell* or *collecting_duct; HormoneStatus* represents positive or negative estrogen, progesterone, or human epidermal growth factor 2 for breast cancer, for example, *er_positive;* and *Laterality* indicates one or both sides of the body, most notably with regard to breast cancer, for example, *bilateral*.

Syndrome and CancerGene

The *Syndrome* and *CancerGene* clusters contain 32 and 141 concepts, respectively. The syndromes included in the ontology are curated directly from the list of syndromes in the ACMG CPG [14], and the cancer genes come from reviewing ACMG and NCCN CPGs as well as genetic tests from well-known cancer testing companies such as Myriad Genetics, ARUP Laboratories, GeneDx, and others. The most important syndromes for evaluating the CPG criteria are LFS and LS along with their associated gene mutations: TP53 for LFS and EPCAM, MLH1, MSH2, MSH6, and PMS2 for LS.

System Architecture

All 192 test cases were built to ensure the accuracy of the implemented criteria completed in 14 minutes and 47 seconds. The longest time a test took to complete was 19.6 seconds, the shortest was 2.9 seconds, and the average was 4.5 (SD 1.9) seconds. The reasoning time varied with the number of family members and the combination of cancers and disease factors present in the FHx. The response time of the entire system (chatbot–report) depends on the number of family members and the total number of cancer diagnoses in the FHx. A typical FHx case with 3 to 5 cancers and 20 to 25 family members takes approximately 20 to 40 seconds. However, various combinations of cancers and family members can take as little as 8 seconds or as long as 5 minutes. The system is asynchronous; it can process FHx for multiple probands at a time (2×CPU count+1 threads) and sends PDF reports to the proband once the reasoner completes and the PDF is rendered. At the end of the assessment, the chatbot notifies the proband that as soon as their FHx is finished processing, the PDF report will be sent to the email address they provided.

In separate studies, we report on proband recruitment and FHx collection [39] and compare the results of ACMG and NCCN criteria applied to the FHx by the system for 4915 probands who have collected their FHx using the system and received a report [52]. Of those, 2221 probands met the criteria, and 2694 did not meet the criteria. Breast and ovarian cancer guidelines were the most consistent, and colorectal and endometrial guidelines were the most disparate across the ACMG and NCCN. Of all probands who did not meet the criteria, 90.6% had cancer in their FHx. In an additional study, we compared the referral patterns for genetic counselors, oncologists, and primary care providers to determine the level of concordance with the CPG criteria implemented by ItRuns [53]. Oncologists and primary care providers had consistently lower rates of concordance with CPG criteria, especially for probands whose FHx triggered the CPG criteria, indicating an immediate opportunity for the system to help frontline care providers identify patients at risk if the system were implemented across the primary care population. Genetic counselors had very high concordance with CPG criteria, especially for probands who met the criteria, and the ontology classification of the system had high concordance with genetic counselors, indicating tight coupling between the CPG recommendations and genetic counselors' professional assessments. The system has strict adherence to CPG criteria and has the potential to reduce human error in FHx collection and risk assessment.

Discussion

Principal Findings

We curated an ontology using Owlready2 and Protégé and developed a web system to apply CPG criteria to patient FHx and identify probands who should consider a cancer genetic consultation. Intersecting the CPG ontology with patient FHx using traditional web development strategies provides patients with access to evidence-based recommendations without requiring the initial time and effort of trained professionals.

Hereditary Cancer CPG Ontology

Potential Impact for Identifying Patients at Risk

Identifying patients at risk of hereditary cancer is a multilayered and highly nuanced challenge. Providers lack time and training for collecting and assessing hereditary cancer risk during patient visits; genetic counselors trained in FHx collection and assessment are in short supply; and patients lack the expertise to interpret CPG criteria for themselves. The end goal is to get patients whose FHx meets the CPG criteria in front of genetic counselors as soon as possible for preventive actions to have the maximum impact on patient outcomes. Chatbots simplify the process of collecting FHx, do not require trained professionals, and are designed for a positive user experience. FHx collected by chatbots is by default in electronic format and ready for analysis. Ontologies are validated tools for modeling CPGs and, with the help of Owlready2, can be accessed using web APIs to assess FHx for hereditary cancer risk. The results can be shared with the patient and the provider before a consultation, effectively removing barriers to referring patients whose FHx indicates hereditary cancer risk to meet with genetic counselors for a cancer genetic consultation.

Owlready2 and Ontology Development

Not all ontologies are naturally adaptable to applying CPG criteria to patient data, and there is certainly ≥1 ontology formalism that would satisfy the needs of the hereditary cancer CPG ontology. Ontologies, especially biomedical ontologies, are generally organized and optimized for dictionary-like

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functions such as looking up information and modeling relationships as closely as possible to the real world. Understandably, this ontology development objective sometimes leads to very large biomedical ontologies, such as Systematized Nomenclature of Medicine–Clinical Terms and the National Cancer Institute Thesaurus, which require impractical processing power for the HermiT Reasoner to reason with. However, more importantly, as these ontologies are not modeled with the patient as a central concept, the HermiT Reasoner is not sufficient to apply CPG criteria out of the box without additional work performed by supporting functions in the FHx API. A lightweight, patient-centric, domain-specific ontology that is small enough to run inside a web API is crucial to quickly apply CPG criteria to FHx. Importantly, a proband's risk depends on whether first-degree relatives meet the CPG criteria. Therefore, each family member in the ontology can be instantiated as an individual of the *Patient* class and be classified according to the CPG equivalency classes.

System Architecture

Domain Ontologies and Service-Oriented Architecture

Lightweight, patient-centric domain ontologies align with a modular service-oriented architecture (SOA) approach to applying CPGs to patient FHx. SOA is a web development architectural pattern that allows small applications to work together over a network to achieve an overall end goal. For example, the chatbot is one such service, and once a patient's FHx is collected, it sends the FHx to the FHx API, which is another service. Once the FHx API has applied CPGs to the patient FHx, it sends the results to another service to create and send the PDF report. The CPG ontology is a component accessed by the FHx API service. As it is lightweight and patient-centric, it can be applied in a modular fashion. For example, the isolation of *ACMGPatient* and *NCCNPatient* in the hierarchy in the ontology (Figure 4) enables a plug-and-play style of applying CPGs to patient FHx. All other classes in addition to *ACMGPatient* and *NCCNPatient* in the hereditary cancer CPG ontology, along with properties and individuals in Figure 4, represent a common set of knowledge needed for evaluating both the ACMG and NCCN criteria. This common set of knowledge between ACMG and NCCN allows *ACMGPatient* and *NCCNPatient* to be executed independently of each other by loading 2 separate, smaller CPG ontologies—an ACMG CPG ontology and an NCCN CPG ontology—that depend on the same domain knowledge. SOA applies these 2 smaller ontologies to patient FHx in parallel and synthesizes their results together at the end, thereby decreasing the time to complete the reasoning with the HermiT Reasoner and sending results to the report service. The system works asynchronously, and the chatbot informs patients that when reasoning is complete, they will be emailed a PDF report with their results. In theory, this approach could be applied to any combination of ontologies for ≥1 domain, as long as the ontologies are sufficiently small and patient-centric.

System Results Compared With Professional Assessments

Automating FHx and hereditary cancer risk assessment reduces irregularities in data collection and the application of CPG criteria. Standardizing FHx collection and the application of CPG criteria is an important step in helping to consistently identify patients at risk of hereditary cancer. It is true that the system is rigidly tied to the CPG criteria, and CPGs are not validated resources. However, CPGs are very valuable, empirically derived benchmarks curated by experts, which could provide an initial screen that is largely currently missing on the frontlines of care. The system is not intended to replace professional assessments but rather complement them. Indeed, we found in our comparison of genetics and non–genetics providers' professional assessment cases that the CPGs applied by the system were discordant with provider recommendations. These cases were often cases where FHx fell just short of meeting a criterion's threshold for cancer cases in the family or age of diagnosis. The initial assessment performed by the system is designed to alert patients and physicians when a professional assessment is warranted strictly according to the CPG criteria. Our preliminary data comparing genetics and non–genetics professionals' concordance with CPGs indicates that non–genetics professionals (primary care physicians and oncologists) unsurprisingly have low concordance, and genetics professionals have high concordance with CPG criteria [53]. The observed high concordance with CPG criteria for genetics professionals and the system is evidence that primary care population-wide application of the system could reduce human error in CPG criteria application to FHx and support primary caregivers in identifying patients at risk of hereditary cancer. In addition, by having formalized actionable rules, cases where providers are discordant with CPGs can be identified as places where the guidelines can potentially be improved. Such improvements could result from human judgment and intuition, which interact with formalized logic encoded within the system.

Importantly, the system is intended to be an initial screen and is not intended to replace professional assessments. In fact, the system is designed to augment the genetics and non–genetics professionals' capacity to collect and assess FHx risk for hereditary cancer by recommending patients whose FHx meets CPG criteria to seek a cancer genetic consultation. Although the system rigidly adheres to nonvalidated CPG guidelines and might at times be discordant with health care providers' professional assessments, the broad application of the system to the primary care population would increase the identification of patients who do meet criteria dramatically from the current state of hereditary cancer risk assessment. Our preliminary data show that genetic counselors have very high concordance with CPG criteria for FHx that does, in fact, meet CPG criteria and non–genetics professionals do not. The notion of false positives and false negatives in this context is nuanced. As the CPGs are not validated tools but are curated by panels of experts, in the case that a health professional is discordant with the CPG, it is difficult to determine who is correct. In the event the system provides evidence-based recommendations for a cancer genetics consultation, and the health care professional disagrees, at least the patient and provider have increased awareness of the patient's FHx risk status for hereditary cancer. In the event the system does not provide an evidence-based recommendation for a cancer genetic consultation, but the provider would recommend counseling, the patient still collects FHx and can show it to their provider in future appointments. In either case, the purpose of the system is fulfilled by collecting FHx and

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applying CPG criteria to assess risk and raise awareness. More work needs to be conducted to implement the system in a broader clinical context; however, strict adherence to CPG criteria would definitely be a step forward from the current lack of application of any FHx risk assessment at the primary care level [52-54].

Limitations

The HermiT Reasoner is a very efficient ontology reasoner and is capable of reasoning over large ontologies. However, even small ontologies take several seconds to reason with, and most web applications typically adhere to subsecond response times for optimal user experience. The computation time required to apply the HermiT Reasoner to patient FHx necessitates an asynchronous experience where patients receive an email with their PDF report 30 seconds to a minute after completing the chatbot questions. Although this is workable, ideally, patients would receive immediate feedback upon completing their FHx collection. We developed a custom rule engine for applying CPG criteria without using the HermiT Reasoner, which increased the processing time substantially. However, more work is required to make it generally applicable across all domains.

Comparison With Prior Work

The prior systems we chose for comparison in this study were systems designed specifically to computerize a wide range of CPGs for broad application and use. The system described in this paper similarly outlines an approach for automating the application of rule-based CPGs. Hereditary cancer was selected to demonstrate the effectiveness of the approach but is intended to be applied to other use cases. There are a number of other technologies designed to encode CPGs for various clinical decision support purposes such as Athena DSS [18], Asbru [19], GEM [20,21], GLIF3 [22,23], EON [24], PROforma [25], GUIDE [26], Prodigy [27], and SAGE [28,29]. However, these systems are older, difficult to access, and more oriented toward integrating with clinical workflows. Clinical workflow integration primarily supports clinicians and is generally less accessible to patients. DESIREE (Decision Support and Information Management System for Breast Cancer) is a more recent example of such a solution that uses ontological reasoning to support CPG application to patients with breast cancer in clinical settings to help tumor boards develop care plans [55-57]. Similar to our system, DESIREE and SAGE used Protégé to develop domain ontologies to computerize CPGs.

A system developed by Abidi [58] has some parallels with our approach. They built a system for a breast cancer follow-up CPG that used GEM to computerize the CPG criteria. Their approach is similar in the use of ontologies to computerize CPGs and execute reasoning to obtain recommendations; however, the methods and applications are quite different. Their system was designed for clinicians to author rules based on the CPGs using GEM, whereas we built a system that outlines a replicable development pattern designed for application in a modern web development environment.

The solution we built is distinctly different in that it applies widely accepted web development best practices to ontology curation and application, focuses on empowering patients with CPG-driven recommendations instead of integrating with provider workflows, and uses a chatbot optimized for mobile devices that simplifies FHx collection and seamlessly interoperates with ontology-driven risk assessment. In addition, our codebase is open source and available on Bitbucket [45].

Conclusions

Combining web APIs, chatbots, ontologies, and hereditary cancer CPGs has the potential to identify patients at risk of hereditary cancer based on patient FHx more efficiently. Patients can collect and receive CPG-driven insights about their FHx before seeing their health care provider, thereby removing the burden of initially collecting and assessing FHx with trained professionals. Ontology-assisted CPG-driven recommendations serve as a temperature check, offering an initial indication of whether patients and providers should consider a cancer genetic consultation based on FHx. Earlier identification of patients at risk for hereditary cancer based on their FHx will result in earlier preventive actions for better outcomes.

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Conflicts of Interest

JBL is the primary developer of Owlready2. HM, JDS, and BMW are the cofounders and shareholders for ItRunsInMyFamily. HM and BMW are the cofounders and shareholders for Dokbot.io.

Multimedia Appendix 1
Clinical practice guidelines and ontology concepts.
[DOCX File , 25 KB - cancer_v8i1e29289_app1.docx]

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Abbreviations

ACMG: American College of Medical Genetics and Genomics
API: application programming interface
CPG: clinical practice guideline
DESIREE: Decision Support and Information Management System for Breast Cancer
EHR: electronic health record
FHx: family health history
LFS: Li–Fraumeni syndrome
LS: Lynch syndrome
NCCN: National Comprehensive Cancer Network
OAO: ontology access object
OPI: ontology programming interface
ORM: object relational mapper
SAGE: Sharable Active Guideline Environment
SOA: service-oriented architecture
SPARQL: SPARQL Protocol and RDF Query Language

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Review

Cancer Screening Recommendations During the COVID-19 Pandemic: Scoping Review

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Abstract

Background: Cancer screening tests are recommended to prevent cancer-associated mortality by detecting precancerous and cancerous lesions in early stages. The COVID-19 pandemic disrupted the use of preventive health care services. Although there was an increase in the number of cancer screening tests beginning in late 2020, screenings remained 29% to 36% lower than in the prepandemic era.

Objective: The aim of this review is to assist health care providers in identifying approaches for prioritizing patients and increasing breast, cervical, and colorectal cancer screening during the uncertainty of the COVID-19 pandemic.

Methods: We used the scoping review framework to identify articles on PubMed and EBSCO databases. A total of 403 articles were identified, and 23 articles were selected for this review. The literature review ranged from January 1, 2020, to September 30, 2021.

Results: The articles included two primary categories of recommendations: (1) risk stratification and triage to prioritize screenings and (2) alternative methods to conduct cancer screenings. Risk stratification and triage recommendations focused on prioritizing high-risk patients with an abnormal or suspicious result on the previous screening test, patients in certain age groups and sex, patients with a personal medical or family cancer history, patients that are currently symptomatic, and patients that are predisposed to hereditary cancers and cancer-causing mutations. Other recommended strategies included identifying areas facing the most disparities, creating algorithms and using artificial intelligence to create cancer risk scores, leveraging in-person visits to assess cancer risk, and providing the option of open access screenings where patients can schedule screenings and can be assigned a priority category by health care staff. Some recommended using telemedicine to categorize patients and determine screening eligibility for patients with new complaints. Several articles noted the importance of implementing preventive measures such as COVID-19 screening prior to the procedures, maintaining hygiene measures, and social distancing in waiting rooms. Alternative screening methods that do not require an in-person clinic visit and can effectively screen patients for cancers included mailing self-collection sampling kits for cervical and colorectal cancers, and implementing or expanding mobile screening units.

Conclusions: Although the COVID-19 pandemic had devastating effects on population health globally, it could be an opportunity to adapt and evolve cancer screening methods. Disruption often creates innovation, and focus on alternative methods for cancer screenings may help reach rural and underresourced areas after the pandemic has ended.

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KEYWORDS

COVID-19; cancer prevention and early detection; cancer screenings; breast cancer screening; cervical cancer screening; colorectal cancer screening

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Introduction

Cancer-associated mortality is the second leading cause of death in the United States [1,2]. Cancer screening tests are recommended to prevent cancer-associated mortality by detecting precancerous and cancerous lesions in early stages [3]. The most common routine cancer screenings include breast, colorectal, and cervical [4].

The COVID-19 pandemic disrupted the use of preventive health care services [5]; there was an abrupt decline in cancer screening services throughout 2020 [6]. A report from May 2020 suggested there was a 94% drop in cancer screening tests across the United States, primarily due to disruptions in access to screening tests [7]. Although there was an increase in the number of cancer screening tests beginning in late 2020, screenings remained 29% to 36% lower than in the prepandemic era [8].

The reduction in cancer screenings and other preventative and diagnostic care have been attributed to both health care provider and patient constraints [9-12]. Health care provider constraints included restrictions on elective procedures [9] and a shortage of health care staff due to redeployment to help with pandemic-related care [9,10]. Even when health care providers have increased availability of preventive care and cancer screenings, many patients face constraints. Patient constraints include loss of income and employer-based insurance coverage [11] and fear of contracting COVID-19 during in-person health care visits [12].

The decline in cancer screening resulted in fewer cancer diagnoses in 2020 [13,14] and raises concerns that missed screenings and delayed cancer diagnoses will likely lead to late stage diagnosis and higher cancer-related mortality [7,14]. For example, a study (n=5167) reported a 13.5% (P=.03) decrease in colorectal cancer diagnoses during March 2020 to December 2020 compared to the number of patients diagnosed before the pandemic, and the same study showed the average number of stage three colorectal cancer cases (advanced stage cancers) diagnosed per month increased by 68.4% (P<.001) [15].

Health care providers must consider ways to increase cancer screening. Therefore, we conducted a scoping literature review to assist health care providers in identifying approaches for prioritizing and increasing cancer screening during the uncertainty of the COVID-19 pandemic. In this review, we focused on the most common cancer screenings: breast, cervical, and colorectal.

Methods

We used the scoping review framework outlined by Arksey and O'Malley [16] to identify and gather evidence from all sources in the field. The framework is comprised of four stages: (1) identification of relevant literature on multiple databases, (2) screening of identified literature and selection of relevant studies, (3) extraction of data, and (4) summarization and reporting of the findings [16]. The research questions of this review are what methods are recommended for risk stratification and triage of patients for cancer screenings, and what alternative cancer screening methods are recommended?

Stage 1: Identification of Relevant Literature

The keywords used to identify articles on PubMed and EBSCO databases were "cancer screening and coronavirus," "cancer screening and COVID-19," and "cancer screening and SARS-CoV-2." The articles selected had to include breast, cervical, and colorectal cancer screening. Articles were screened for relevance based on the information provided in the abstract, and those deemed to be relevant by their abstract were fully reviewed. Additional literature was identified from the references of selected articles. A broader search strategy was adopted to include gray literature. These included commentaries and editorials published in peer-reviewed journals, recommendations published by professional organizations or societies, and medical news articles. The literature review ranged from January 1, 2020, to September 30, 2021.

Stage 2: Screening of Identified Literature and Selection of Relevant Studies

A total of 350 articles were identified from the databases, and an additional 53 articles were identified from references of the relevant articles. After pooling literature from different sources, we found 192 articles were duplicates; duplicates were excluded. Of the remaining 211 articles, 168 were deemed ineligible after screening the abstracts. Of the remaining 43 articles that were fully reviewed, 20 were excluded. Articles not focused on breast, cervical, and colorectal cancer screenings; not suggesting measures to address cancer screening during and after the pandemic; and providing suggestions not substantiated by past literature were excluded. A total of 23 articles were selected for this review. Two authors (SKS and PAM) reviewed the literature and agreed upon the selection of articles. The PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flowchart is provided in Figure 1.

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Figure 1. PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow diagram for new systematic reviews, which includes searches of databases only.

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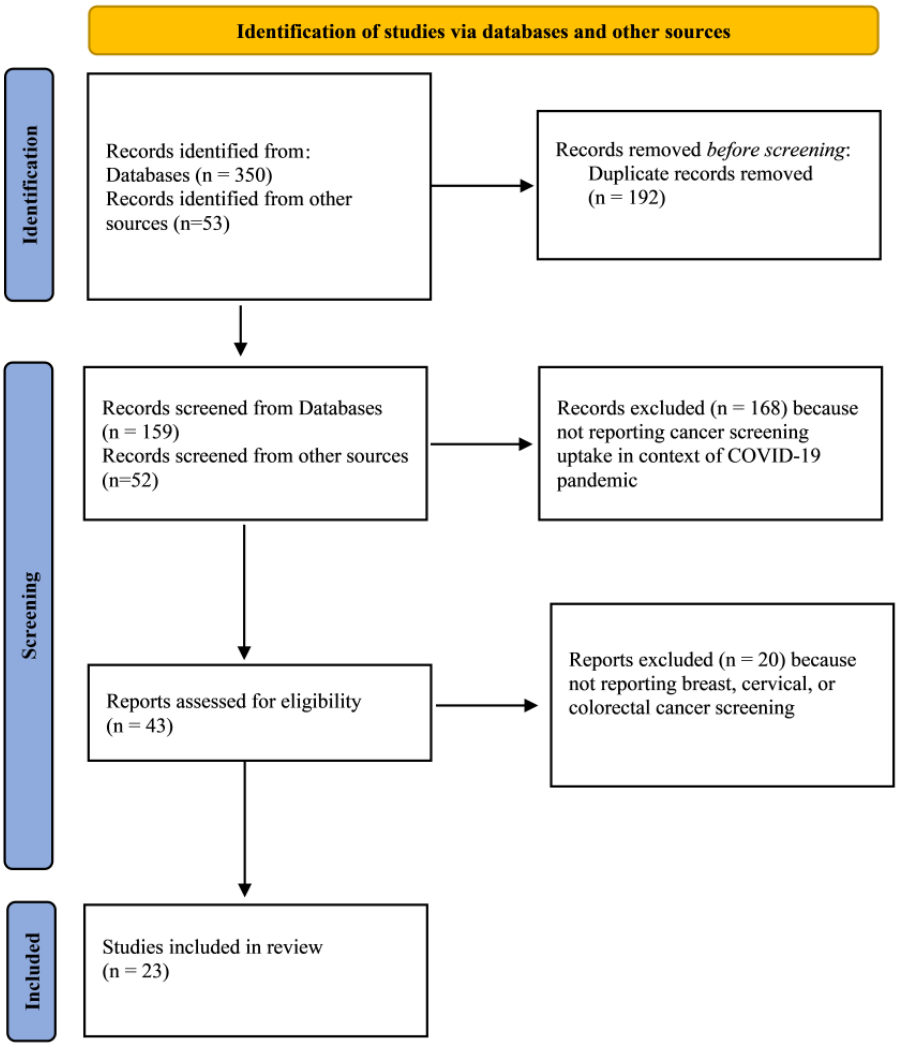

Stage 3: Extraction of Data

The data points recorded were the article citations, type of article, type of cancer screening discussed, and key recommendations.

Stage 4: Summarization and Reporting of the Findings

The Results section and tables summarize the data regarding recommendations for risk stratification and triage and alternative cancer screening methods for breast, cervical, and colorectal cancer screenings and report concise information about alternative methods that can be used for cancer screenings.

Results

The articles included two primary categories of recommendations: (1) risk stratification and triage to prioritize screenings and (2) alternative methods to conduct cancer screenings (Table 1).

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Table 1. Summary of safely resuming cancer screening services.

Approach	References	Strategies
Risk stratification and triage	• Basu et al 2021 [9] • Castanon et al 2021 [17] • Cohen et al 2020 [18] • Corley et al 2021 [19] • Croswell et al 2021 [6] • Fagundes et al 2021 [20] • Gralnek et al 2020 [21] • Helsper et al 2020 [22] • Houlihan 2020 [23] • Isaacs and Leininger 2021 [24] • Issaka and Somsouk 2020 [25] • Kadakuntla et al 2021 [26] • Miller 2021 [27] • Orenstein 2020 [28] • Pediconi et al 2020 [29] • Puricelli Perin et al 2021 [30] • Riley 2020 [31] • Seguin 2020 [32]	• Stratify patients into high-risk, average-risk, and low-risk categories based on age, sex, past medical history, past personal history, or region/area of residence • Triage patients based on risk category, prioritizing patients at high risk of cancer, followed by average-risk and low-risk patients
Alternative screening methods	• Balzora et al 2020 [33] • Castanon et al 2021 [17] • Corley et al 2021 [19] • Croswell et al 2021 [6] • Fagundes et al 2021 [20] • Gorin et al 2021 [34] • Issaka and Somsouk 2020 [25] • Kadakuntla et al 2021 [26] • Miller 2021 [27] • Miller et al 2021 [35] • Orenstein 2020 [28] • Ricciardiello et al 2021 [36]	• Self-collecting of vaginal or urine samples for cervical cancer screening • Self-collection of stool sample for colorectal cancer screening • Mobile units outside primary health care facilities for breast cancer screening

Risk Stratification and Triage

Risk stratification and triage was recommended as an appropriate method for focusing cancer screenings during the COVID-19 pandemic. The recommendation focused on prioritizing those who are most susceptible to developing cancers [6,9,17-32]. Potential criteria considered for categorizing patients into high risk included patients with an abnormal or suspicious result on the previous screening test [27], age group [17,26,32], sex [26], personal medical or family history [18,24,26,27], currently symptomatic or asymptomatic, predisposition to hereditary cancers, and inheritance of cancer-causing mutations [18,26].

Conversely, articles recommended the following patients be deferred until high priority patients are offered cancer screenings: patients with a recent cancer screening with normal results [17,20]; patients who do not have any cancer-related symptoms [18,22]; patients who have taken prophylactic measures such as the human papillomavirus (HPV) prophylactic vaccine [17,20,24]; and patients who do not have medical, personal, or family-related indication for immediate screening [18,19,23,31].

Other recommended strategies included identifying areas facing most disparities [19,34], creating algorithms [24] and using artificial intelligence [28] to create cancer risk scores, leveraging in-person visits to assess cancer risk [6], and providing the option of open access screenings where patients can schedule screenings and can be assigned a priority category by health care staff [26]. Some recommended screening high-risk patients through telemedicine prior to having them come into health care providers [23,29].

In addition to risk stratification and triage, telemedicine was recommended to determine screening for patients with new complaints [18,19,22]. Several articles noted the importance of implementing preventive measures such as COVID-19 screening prior to the procedures [6,9,26], maintaining hygiene measures [19,32], and social distancing in waiting rooms [32].

Alternative Screening Methods

Several studies discussed using novel and alternative screening methods that do not require an in-person clinic visit and can effectively screen patients for cancers (Table 2). Mailing of self-collection sampling kits was widely suggested as a screening strategy for cervical and colorectal cancers [6,17,19-21,25-28,33-36]. Cervical cancer screening included mailing or pharmacy pickup of kits for self-sampling of vaginal or urine samples that can be tested for HPV strains most likely to cause cancers [17,27,34,35]. Stool-based self-collection kits that are performed at home and mailed for screening were recommended for colorectal cancers [6,19,20,25,26,33-36]. Although self–breast examinations can be done at home, they do not take the place of mammography; therefore, articles recommended implementing and expanding mobile screening units [28,30].

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Table 2. Alternative approaches to increase cancer screenings.

Cancer type/cancer risk factors	Conventional recommendation/practices	Variation in approaches
Breast cancer	• Mammography	• Screening at mobile units or small satellite units • Follow-up on patients with abnormal results
Cervical cancer	• Pap smear • Pap smear + HPV[a] co-testing	• Self-collection of vaginal or urine samples at home • Follow-up on patients with abnormal results
Colorectal cancer	• Colonoscopy • Sigmoidoscopy • CT[b] colonography • Stool-based tests	• Self-collection of stool samples at home • Follow-up on patients with abnormal results

[a]HPV: human papillomavirus.

[b]CT: computed tomography.

Discussion

The number of cancer screenings missed during the COVID-19 pandemic will likely lead to a sharp increase in the number of late-stage cancer diagnoses and increased cancer mortality [14]. As health care providers look for ways to focus their cancer screening efforts, this review provides insights into risk stratification and triage approaches and alternative screening approaches that can be adopted to reduce the impact of COVID-19 on cancer mortality.

Risk stratification and triage approaches focused on prioritizing patients based on personal characteristics, medical history, cancer screening history, and communities facing highest cancer disparities [6,9,17-32]. The literature suggests that older patients at higher risk should be given priority since the risk of cancer increases with age [17,26].

Prioritizing high-risk patients based on the past screening history could help the health care provider prioritize care based on the probability of patients developing cancerous lesions. Several studies have shown that prioritizing high-risk patients based on past medical history is important [6,9,17-32], and studies have reported the effectiveness of the personalized screening approach, demonstrating that the one-size-fits-all approach may not be the best strategy [37-40]. In addition, using algorithms and artificial intelligence to categorize and triage high-risk patients will help navigate large data sets and assist physicians in the decision-making process [24,28].

Alternative cancer screening approaches focused on tests that do not require a clinic or hospital visit can be used to collect samples at home. These alternative methods allow initial screening outside the traditional clinical environment, take fewer clinical resources, and reduce exposure risk to patients. Alternative at-home screening modalities exist for cervical cancer screening [41-43] and colorectal cancer [26]. Studies have evaluated the efficacy of detecting cervical intraepithelial lesions using self-collected samples with samples collected in the doctor's office and concluded that self-sampling is a safe and effective alternative to screen for cervical cancers [42,43]. Similar to cervical cancer, colorectal cancer screenings can be effectively conducted using noninvasive stool-based test kits at home [44,45]. Studies have shown that stool-based test kits can help reach underresourced communities and increase colorectal cancer screening uptake [46]. Although the stool-based tests have a high false-positive rate [47], patients testing negative can be assured that they do not have colorectal cancers [26].

Follow-up for abnormal results from at-home tests can be provided and help focus limited clinical resources. Although there are not at-home alternatives for mammography, mobile units can provide a way to reach the community [28,30] and reduce exposure risk.

Although the COVID-19 pandemic had devastating effects on population health globally, it could be an opportunity to adapt and evolve our cancer screening recommendations. Disruption often creates innovation, and focus on alternative methods for cancer screenings may help reach rural and underresourced areas after the pandemic has ended.

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Conflicts of Interest

None declared.

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Multimedia Appendix 1
PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) checklist.
[DOCX File , 31 KB - cancer_v8i1e34392_app1.docx]

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Abbreviations

HPV: human papillomavirus
PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses

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Review

Remote Psychological Interventions for Fear of Cancer Recurrence: Scoping Review

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Abstract

Background: Patients with cancer and survivors may experience the fear of cancer recurrence (FCR), a preoccupation with the progression or recurrence of cancer. During the spread of COVID-19 in 2019, patients and survivors experienced increased levels of FCR. Hence, there is a greater need to identify effective evidence-based treatments to help people cope with FCR. Remotely delivered interventions might provide a valuable means to address FCR in patients with cancer.

Objective: The aim of this study is to first discuss the available psychological interventions for FCR based on traditional cognitive behavioral therapies (CBTs) or contemporary CBTs, in particular, mindfulness and acceptance and commitment therapy, and then propose a possible approach based on the retrieved literature.

Methods: We searched key electronic databases to identify studies that evaluated the effect of psychological interventions such as CBT on FCR among patients with cancer and survivors.

Results: Current evidence suggests that face-to-face psychological interventions for FCR are feasible, acceptable, and efficacious for managing FCR. However, there are no specific data on the interventions that are most effective when delivered remotely.

Conclusions: CBT interventions can be efficacious in managing FCR, especially at posttreatment, regardless of whether it is delivered face to face, on the web, or using a blended approach. To date, no study has simultaneously compared the effectiveness of face-to-face, web-based, and blended interventions. On the basis of the retrieved evidence, we propose the hypothetical program of an intervention for FCR based on both traditional CBT and contemporary CBT, named Change Of Recurrence, which aims to improve the management of FCR in patients with cancer and survivors.

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KEYWORDS

fear of cancer recurrence; cognitive behavioral therapy; acceptance and commitment therapy; mindfulness; eHealth; blended intervention

Introduction

Fear of Cancer Recurrence

Along the trajectory of cancer care, which in recent years has been extended because of new technologies [1], patients and survivors might experience fear of cancer recurrence (FCR), which is the concern that cancer will progress when it is stable or that it may return after the end of treatment [2]. Low levels of FCR could be adaptive for patients and survivors, as they promote and encourage the maintenance of medical follow-up, engagement in healthy lifestyle changes, and greater attention to signs of new or recurring cancer [3]. However, when FCR becomes distressing, patients can engage in negative health behaviors (eg, overusing health services and avoiding appropriate tests to identify cancer recurrence) and experience

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higher psychological distress and poorer quality of life (QoL) [3-5]. In addition, significant relationships emerged between FCR and certain sociodemographic variables (eg, younger age and having young children) and psychological factors (eg, anxiety and depression), whereas conflicting data emerged regarding the characteristics of tumor and cancer treatments [3]. With the advent of emerging nanomaterial-based approaches that would promote early diagnosis [6], these could favor better treatment efficacy and a reduction in FCR. However, this is an innovative area that is yet to be explored.

Currently, patients with cancer and survivors experience higher levels of loneliness, FCR, anxiety, and depression because of the diffusion of COVID‐19. Indeed, the pandemic forced people to social distance and caused several challenges for patients with cancer and survivors, such as maintaining social support and continuing their cancer treatment or medical check-ups regularly [7-10]. Despite the current situation, the need to identify effective evidence-based treatments to help people cope with FCR has increased in the past decade. In clinical settings, clinicians can help patients and survivors using psychological support, psychoeducational interventions, or psychotherapy. Data from recent systematic reviews and a meta-analysis of 23 controlled trials have shown that psychological interventions for FCR are feasible, acceptable, and efficacious for managing FCR, having a small-to-medium but a robust effect that persists at postintervention (Hall et al [11]: Hedge g=−0.36; Tauber et al [5]: Hedge g=0.33) and follow-up (Tauber et al [5]: Hedge g=0.30) [4,12]. Interestingly, in these reviews, most psychological interventions were based on traditional cognitive behavioral therapies (CBTs) or contemporary CBTs (eg, mindfulness and acceptance and commitment therapies [ACTs]). The first, traditional CBTs, focused on the contents of thoughts and aimed to identify and modify people's negative thoughts or biases to reduce dysfunctional emotions and promote psychological adjustment, whereas the second, contemporary CBTs, focused on mental processes and aimed to modify how people relate to their inner experiences [3-5,11,12]. In particular, Tauber et al [5] found that FCR symptoms were more responsive to contemporary CBTs (Hedge g=0.42) compared with traditional CBT (Hedge g=0.24) at postintervention; however, this greater effect did not persist over time [5]. Considering the existence of several declinations of CBT approaches, it is important to understand the interventions that are effective in helping patients with cancer with decreasing FCR. As the pandemic forced patients with cancer into obliged isolation, a critical analysis of the different modalities in which these types of therapies can be delivered (eg, face to face or via remote and web-based approaches) might be useful. Indeed, apart from traditional in-presence interventions, blended (a mixed method comprising both web-based and face-to-face therapy) or remote care for patients with cancer has been increasingly applied [13]. Furthermore, during the pandemic, the management of patients with cancer has raised medical issues [14,15] and psychological issues [16].

Objective

In this paper, we aim to critically revise and systematize the available evidence on the effectiveness of different modalities and approaches of CBT psychological interventions for FCR. Specifically, this review aims to summarize studies dealing with face-to-face, remote, and blended interventions based on traditional CBTs or contemporary CBTs, in particular, mindfulness and ACT, used to reduce FCR. Finally, we propose a possible program based on the retrieved literature.

Methods

Electronic searches were performed using PubMed, MEDLINE, and Embase between November and December 2020, with no time limits. Original articles were considered in English, Italian, or Spanish languages, with participants aged ≥18 years. Keywords searched in titles and abstracts included *fear of cancer recurrence* combined with terms such as *cognitive behavioral therapy*, *acceptance and commitment therapy*, *mindfulness*, *mindfulness-based stress reduction*, *blended therapy*, *face-to-face intervention*, and *online intervention*. The reference lists in the relevant systematic reviews were manually searched for additional contributions that met our inclusion criteria. The search was limited to only full-text articles. The studies included in this paper met the following main criteria: (1) articles dealing with patients with cancer with FCR, (2) articles including traditional or contemporary cognitive psychological interventions for FCR (CBT, ACT, and mindfulness), and (3) articles presenting quantitative data or study protocols. Commentaries, editorials, and conference proceedings were excluded. Research papers on traditional and contemporary CBT interventions (classical CBT, mindfulness, and ACT) were included.

Initially, the search strategy yielded 470 articles that were screened for irrelevant or duplicate articles. The remaining articles were assessed and selected by screening the abstracts, followed by full-text reading and selection according to the predefined inclusion and exclusion criteria. Of the 470 articles, 35 (7.4%) articles that focused on psychological interventions for reducing FCR were selected. The results were organized according to the modality of delivery of the therapy (face to face, remote, and blended).

Results

Face-to-face Traditional CBT, Mindfulness, and ACT

Psychological interventions for patients with cancer and survivors based on the principles of traditional CBT aim to encourage patients to identify, express, and deal with their fears and emotional reactions related to cancer and improve their ability to cope with them to maintain their QoL and evaluate and alter life priorities [17-23]. Traditional CBT interventions can be delivered in groups [17,19,21,22,24] or individually [18,20,23,25] and have short-term benefits [17,19,22-24] and significant long-term effects on FCR [17,18,20].

Clinicians often begin the interventions with psychoeducation on FCR to explain what it is and how it presents and maintains during everyday life [22,23]. Moreover, through these interventions, patients can improve their problem-solving skills and use of personal and social resources, enhance their self-esteem and autonomy [20,21], and look into their illness

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beliefs and behaviors [18]. Specifically, patients address irrational thoughts through cognitive reframing to reduce catastrophic interpretations of physical symptoms and emotions [19,21-23]. Some examples of psychological interventions are Side by Side, a couple of skill interventions proposed by Heinrichs et al [24], and the adjustment to the fear, threat or expectation of recurrence intervention theorized by Humphris and Rogers [18]. In particular, the Side by Side program takes place at the couple's home and comprises 4 biweekly face-to-face sessions with a therapist, focused on communication skills and dyadic coping [24]. Instead, the adjustment to the fear, threat or expectation of recurrence intervention is for patients—the caregiver can only be included if desired by the patient—and is intended to encourage patients to express and process fears related to FCR and explore illness beliefs and behaviors. During the sessions, patients also practiced relaxation exercises [18].

Psychological interventions based on mindfulness, which is a particular way to pay attention to the present moment without judgment, ensure that people turn away from unhealthy beliefs, thoughts, or emotions, maintaining awareness of the present moment [26]. Clinicians can choose between different mindfulness-based programs, such as mindfulness-based stress reduction (MBSR), mindful movement program, or mindfulness-based cognitive therapy (MBCT) [4,5,27]. The MBSR program aims to self-regulate arousal in response to stressful situations or symptoms and reduce the intensity of cognitive processes by lowering the frequency of negative automatic thinking [4,27]. This program comprises 1-hour session conducted by a psychologist and home practice following a manual and audiotapes (eg, sitting meditation, walking meditation, body scan, and yoga), with data showing that it has a significant effect on FCR in people with cancer at postintervention [28-32]. Instead, the mindful movement program combines mindfulness with self-directed movement for patients to explore and understand their thoughts, feelings, and sensations. Data report a significant effect on FCR at 12 weeks after treatment; however, this effect does not maintain at 18 weeks after treatment [33]. The MBCT program aims to teach participants to be more mindful in daily life through meditation exercises, yoga, group discussions, and didactic teaching. This program comprises 8 weekly sessions, a silent day, and daily homework. Compared with usual care, patients who receive this program report significantly lower levels of FCR (Cohen d=0.27) [27].

Finally, ACT interventions explain psychological distress through psychological inflexibility, a construct that comprises behaving under the strict control of rigid personal thoughts, feelings, and other internal experiences [34]. ACT aims to reduce psychological inflexibility, limit the use of maladaptive coping strategies, and enhance psychological flexibility, which lets people live mindfully according to their values and accept both negative and positive events [34]. However, in the literature, there were only 9% (3/35) of studies that applied ACT to FCR [35-37], and only 3% (1/35) was a randomized controlled trial (RCT) assessing the feasibility and preliminary efficacy of ACT interventions for FCR at postintervention and over time [36]. Johns et al [36] proposed a group intervention based on ACT principles, which aimed to promote adaptive coping through acceptance, cognitive defusion, awareness, and perspective-taking exercises while supporting survivors of breast cancer (BC) in aligning their behavior with personal values to cope with fears. Furthermore, participants improved the skills learned during the session by performing home awareness practice [36]. Arch and Mitchell [35] developed a group manual and workbook for participants comprising experiential exercises, metaphors, discussions, and homework. This was aimed at helping participants to be aware of and accept thoughts and emotions about cancer, eliminate rigid thoughts and beliefs about cancer and themselves through psychological flexibility, and define personal values and commit to pursuing meaningful activities in line with those values [35]. All these interventions suggest that ACT intervention could be useful for reducing FCR [35-37]. Moreover, in the literature, a manualized intervention, called ConquerFear, focuses on reducing the impact of FCR based on metacognitive therapy, the Common Sense Model of illness, the Self-Regulation of Executive Function Model, and relational frame theory, which form the theoretical basis for ACT (more details are provided in Butow et al [38]). In particular, this intervention comprises 5 face-to-face sessions, each of which is associated with home exercises on the skills learned in the session. Patients, through the ConquerFear intervention, learned new strategies to control worry, modified dysfunctional beliefs related to worry, acquired appropriate monitoring and screening behaviors, learned to accept the uncertainty caused by a cancer diagnosis, and defined values and goals based on them. This intervention reduced FCR severity at posttreatment and over time [39,40]. A summary of the described face-to-face psychological interventions for FCR is provided in Table 1.

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Table 1. Face-to-face psychological interventions.

Therapy and authors	Cancer	Study design	Intervention and groups	Results
Traditional CBT [a]				
Heinrichs et al [24]	BC[b] or GC[c]	RCT[d]	• Side by Side: 4 biweekly couple skills sessions • Couples Control Program	Side by Side had a greater effect on FoP[e] than the Couples Control Program; however, this difference disappeared by 16 months after the diagnosis.
Herschebach et al [17]	Different type	Longitudinal study	• 4 session cognitive behavioral group therapy • Supportive experimental group therapy • UC[f]	FoP decreased significantly over time in both intervention groups in contrast to the control group that showed only short-term improvements.
Humphris and Rogers [18]	Different type	RCT	• AFTER[g]: 6 weekly sessions of traditional CBT individual therapy • UC	AFTER intervention improves FCR[h] only at the immediate short-term follow-up (MWU[i]: z=2.06; *P*=.04).
Lebel et al [19]	Survivors of BC or ovarian cancer	A single-arm multi-site study	• 6-week cognitive-existential group intervention	Significant reductions of FCR levels immediately after it and at the 3-month follow-up
Manne et al [20]	GC	RCT	• CCI[j]: 7 weekly sessions of individual therapy and 1 telephone session 2 or 3 weeks after session 7 • A supportive counseling intervention: 7 weekly sessions of individual therapy and 1 telephone session 2 or 3 weeks after session 7 • UC	CCI did not affect FCR.
Savard et al [22]	Different type	Development and feasibility	• 4 weekly group CBT sessions	Significant reductions of FCR levels
Tomei et al [23]	Different type	RCT pilot	• FCR intervention: 6-week sessions	Significant reduction of FCR levels at postintervention and at 3-month follow-up
Contemporary CBT				
Crane-Okada et al [33]	Survivors of BC	RCT pilot	• 12-week mindful movement program intervention	Significant effect on FCR at 12 weeks posttreatment; however, this effect does not maintain at 18 weeks after treatment
Lengacher et al [28]	Survivors of BC	RCT	• 6 - week MBSR[k] program • UC	MBSR reduces FCR more than usual care (11.6 vs 9.3) at 6 weeks.
Lengacher et al [29]	Survivors of BC	Feasibility of the intervention	• 8-week MBSR program	Significant effect on FCR
Lengacher et al [30]	BC	A single-arm multi-site study	• 6-week MBSR (BC) program • UC	MBSR (BC) reduces FCR; MBSR (BC) compared with UC had a favorable change in FCR problems that mediated the effect of MBSR (BC) on 6-week change in perceived stress (z=2.12; *P*=.03) and state anxiety (z=2.03; *P*=.04)
Lengacher et al [31]	Survivors of BC	RCT	• 2-hour sessions once per week for 6 weeks of an MBSR (BC) • UC	Significant improvement of FCR in the MBSR (BC) group compared with usual care
Victorson et al [32]	PC[l]	RCT pilot	• 8 - week MBSR intervention • An attention control arm	MBSR significantly reduces PC anxiety and uncertainty intolerance

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Therapy and authors	Cancer	Study design	Intervention and groups	Results
Arch and Mitchell [35]	Survivors of BC	Pilot study	• 7 weekly 2-hour sessions of ACT[m]	FCR decreases through 1 week following the last group session (post; Cohen *d*=0.34; *P*<.05) and 3 months following post (Cohen *d*=0.66; *P*=.001) but not during the month-long baseline period (Cohen *d*=0.11; *P*=.43)
Johns et al [36]	Survivors of BC	RCT	• 6 weekly 2 - hour group sessions of ACT • 6 weekly 2 - hour group sessions of SE[n] • One 30 - minute group coaching session (EUC[o])	SE and ACT reduce FCR severity over time; however, only ACT produced significant reductions at each time point relative to baseline, with between-group differences at time point 4 substantially favoring ACT over SE (Cohen *d*=0.80; *P*<.001) and EUC (Cohen *d*=0.61; *P*<.01)
Marín and Soriano [37]	BC	Open trial	• 1 session of ACT • Waiting list	Defusion contributes to decreasing FCR, and this effect is maintained 3 months after the intervention.
Butow et al [38,39]	BC or CRC[p]; BC or CRC or melanoma	Study protocol of an RCT; RCT	• ConquerFear intervention: 5 face-to-face sessions over 10 weeks • Taking-it-Easy relaxation therapy: 5 face-to-face sessions over 10 weeks	ConquerFear is efficacy compared with attention control in reduction of FCR immediately after therapy and 3 and 6 months later
Smith et al [40]	BC	Pilot study	• ConquerFear intervention: 5 face-to-face sessions over 10 weeks	ConquerFear is feasible, acceptable, and shows potential efficacy for FCR

[a]CBT: cognitive behavioral therapy.
[b]BC: breast cancer.
[c]GC: gynecologic cancer.
[d]RCT: randomized controlled trial.
[e]FoP: Fear of Progression.
[f]UC: usual care.
[g]AFTER: adjustment to the fear, threat or expectation of recurrence.
[h]FCR: fear of cancer recurrence.
[i]MWU: Mann-Whitney *U* test.
[j]CCI: coping and communication-enhancing intervention.
[k]MBSR: mindfulness - based stress reduction.
[l]PC: prostate cancer.
[m]ACT: acceptance and commitment therapy.
[n]SE: survivorship education.
[o]EUC: Enhanced Usual Care.
[p]CRC: colorectal cancer.

All these approaches, which are delivered face-to-face, present some criticisms: they were relatively expensive and time and resource intensive, and patients could be reluctant to return to the hospital where cancer treatment took place [41]. In the current period, such limits might be overcome by remotely delivered internet-based interventions.

Traditional CBT, Mindfulness, and ACT Delivered Through eHealth

Over the past decades, owing to the increasing use of new technologies for the treatment of psychological aspects, clinicians have applied remotely delivered psychological techniques in the field of mental health and health care settings, giving rise to eHealth [42]. Web-based interventions overcame the criticisms of face-to-face interventions, as they can be performed from patients' homes, even for those who are in remotely located places or have reduced mobility for health issues [43,44]. Moreover, eHealth reduces health care costs, as it requires few economical resources to be allocated for personnel needed compared with the need for personnel's engagement in face-to-face therapies [45]. Finally, eHealth also offers the chance for self-management and continuity of care [46]. However, there are some disadvantages related to eHealth interventions, such as the lack or reduction of personal interaction, poor adherence, and less engagement [47].

Considering the clinical target, eHealth proved to be effective in the field of mental health and psychological treatments [42,48], whereas in specific oncological settings, it has limited and inconsistent findings [49,50].

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eHealth based on CBT involves the delivery of clinical CBT content via the internet and provides content in several formats, for example, text, video, and audio files and interactive elements.

Regarding traditional CBT, Lichtenthal and al [51] proposed a home-delivered cognitive bias modification intervention to reduce FCR. The intervention, Attention and Interpretation Modification for Fear of Breast Cancer Recurrence (AIM-FBCR), targets 2 types of cognitive biases: attentional bias, which is assessed through a modified dot-probe task, and interpretation bias, which is assessed through a word–sentence association paradigm [51]. In a preliminary pilot randomized controlled study (n=110), participants completed 8 personalized treatment sessions twice a week for 4 weeks, and the results showed that this intervention was effective in reducing health worries in patients with cancer [51]. Another example is the web-based self-help training performed by van Helmondt et al [52]. The program, Cancer Recurrence Self‐Help Training trial, comprises a psychoeducation model and a model based on the basic principles of CBT, and 4 optional modules (rumination, action, relaxation, and reassurance) to choose from. Each module comprises an instructive part (texts, videos, or audio files) and a part made of exercises. Patients could be supported on the web personally by emailing a coach [52]. However, the same authors found that there was no effect of this intervention on FCR [44].

Regarding contemporary CBT, specifically for the concerns regarding eHealth mindfulness-based programs used alone in the cancer settings, evidence related to FCR is scarce. However, the internet-based MBCT (eMBCT) intervention, which is a combination of MBCT and CBT, demonstrated reductions in FCR (Cohen d=0.53) and rumination and improvements in mental health–related QoL compared with standard care [27]. Patients accessed this intervention, which was delivered individually, through a secure website in which they found an introduction module and daily meditation exercises with meditation audio files. During the intervention, the patients had to fill out practice diaries. The intervention lasted 8 weeks and included a silent day, at the end of which patients were asked to write about their experiences in an essay and send it to the therapist, who returned written feedback [27]. On the basis of MBSR, there is a mobile MBSR for BC program proposed by Lengacher et al [53]. Patients accessed this program via an iPad after having received a user manual and orientation on how to use the iPad. The content of the program comprised video files on formal meditative techniques (eg, sitting meditation, walking meditation, body scan, and gentle Hatha yoga) and audio files on informal meditative techniques (eg, integrating mindfulness into daily life activities). Moreover, participants could learn the formal and informal meditative techniques provided on the iPad through a physical manual. The results of a pilot study on this program showed that it might be feasible and acceptable, improving FCR scores [53].

Finally, with regard to web-based interventions based on ACT, the studies recovered in the literature were few, and in the oncology field, we found only 11% (4/35) of studies; 50% (2/4) of them were for patients with cancer and the others for partners [54-57]. One of the studies explored the acceptability, feasibility, efficacy, and cost-effectiveness of an internet intervention for survivors of BC called iNNOVBC [57]. To date, the authors have presented only the protocol of this intervention, which comprises 10 modules, 5 of which are optional. In the mandatory modules, there was 1 about anxiety, worries, and fear of recurrence; for that reason, we presented this program. The key components were psychoeducation; 4 of the 6 core processes of psychological flexibility (acceptance, cognitive defusion, values, and committed action); behavioral activation; and relaxation that patients learned through experiential exercises, metaphors, and homework. During therapy, patients could communicate with a therapist via SMS text messaging, chat, email, and videoconference [57].

Finally, in the literature, we found examples of web-based interventions based on multiple theories, such as e-TC and iConquerFear [49,58]. The e-TC includes 6 interactive modules that take approximately an hour to complete. The therapeutic contents were based on traditional CBT, ACT, metacognitive therapy, mindfulness, and relaxation. Patients were trained to restructure or accept their unpleasant thoughts and feelings and cope with stressful situations [49]. The results of the pilot tests showed that the participants were highly satisfied with the program, although they suggested that limited time for men was an obstacle to using and completing the program, and men with a more recent diagnosis and a level of higher distress may be more likely to commit to the program. However, e-TC appears to be a feasible and acceptable web-based intervention for survivors of testicular cancer [49]. To date, no data have supported the effectiveness of this intervention. Moreover, the iConquerFear intervention, based on the aforementioned ConquerFear therapy manual [38], comprises 6 modules, 5 therapeutic and 1 as the introduction, including audio, video, and text contents. Patients completed the modules in 1 to 2 hours over 1 to 2 weeks, and between the modules, they practiced with the skills learned [58]. However, no data supported the effectiveness of this intervention, although there is only a qualitative evaluation of the usability of iConquerFear [58].

A summary of the aforementioned studies on psychological interventions delivered through eHealth is provided in Table 2.

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Table 2. Remote psychological interventions.

Therapy and authors	Cancer	Study design	Intervention	Results
Traditional CBT[a]				
Lichtenthal et al [51]	BC[b]	RCT[c] pilot	• AIM - FBCR[d]: 8 personalized treatment sessions of 30 minutes each administered twice a week for 4 weeks • A control condition program	The results of the current pilot study suggest the promise of AIM - FBCR in reducing FCR[e] in survivors of BC
van Helmondt et al [44,52]	Survivors of BC	Study protocol; RCT	• The Cancer Recurrence Self - help Training trial: less fear after cancer—a tailored web-based self-help training (2 basic modules and 4 optional modules) • UC[f]	There was no effect of the CBT - based web-based self - help training *Less fear after cancer* on FCR in the study
Contemporary CBT				
Compen et al [27]	Different type of cancer	RCT	• Individual internet-based MBCT[g]: access to a secure website containing material for 8 weeks plus a silent day and an inbox+weekly asynchronous written interaction with a therapist over email • MBCT: 8 weekly 2.5-hour group sessions, a 6-hour silent day, and daily home practice assignments guided by audio files • UC	Compared with UC, both interventions reduced FCR
Lengacher et al [53]	BC	Feasibility	• mMBSR (BC)[h]: sitting and walking meditation, body scan, and yoga and is designed to deliver weekly 2 - hour sessions for 6 weeks using an iPad	There was a significant improvement from baseline to 6 weeks after mMBSR (BC) in FCR
Mendes-Santos et al [57]	Survivors of BC	Study protocol	• iNNOVBC[i]: a 10-week guided internet-delivered individually tailored ACT[j] influenced CBT intervention • UC	Not yet available
Heiniger et al [49]	TC[k]	Pilot study	• e-TC: 6 interactive modules for 10 weeks	e-TC appeared to be a feasible and acceptable web-based intervention for survivors of TC
Smith et al [58]	Different type of cancer	Study protocol	• iConquerFear: 5 therapeutic modules completed in 1 to 2 hours over 1 to 2 weeks	Not yet available

[a]CBT: cognitive behavioral therapy.

[b]BC: breast cancer.

[c]RCT: randomized controlled trial.

[d]AIM - FBCR: Attention and Interpretation Modification for Fear of Breast Cancer Recurrence.

[e]FCR: fear of cancer recurrence.

[f]UC: usual care.

[g]MBCT: mindfulness-based cognitive therapy.

[h]mMBSR (BC): mobile mindfulness - based stress reduction for breast cancer.

[i]iNNOVBC: a guided internet-delivered individually tailored acceptance and commitment therapy–influenced cognitive behavioral intervention to improve psychosocial outcomes in breast cancer survivors.

[j]ACT: acceptance and commitment therapy.

[k]TC: testicular cancer.

Traditional CBT, Mindfulness, and ACT Through Blended Care

Currently, in the field of eHealth treatments, the use of blended care is gaining rising visibility. Blended treatment or blended care are defined in literature as "technology-supported care," with the term *blended* describing a combination of web-based and offline elements inside the same care-flow intervention. Generally, the offline part corresponds to face-to-face sessions, whereas for the web-based element, patients accessed a website or workbook, which facilitated further skill acquisition and learning [41]. To date, a single definition of what blended

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intervention exactly reserves was absent, as blended care is currently built with different approaches across studies [48].

In blended treatments, web-based and offline components are not standalone treatment pathways but rather interrelated methods that are strategically combined to build an intervention that merges the potential benefits of the 2 approaches [48,59]. In particular, blended care shares with eHealth the advantage of being flexible in application, having good accessibility, and saving travel time and costs [59,60]. It shares with face-to-face interventions the benefit of having a therapist as a guide, which increases adherence, prevents dropout, facilitates increased treatment intensity, and leads to better results than unguided treatments [61]. To date, the literature has demonstrated that blended therapy displays encouraging effects [48,62] and has shown efficacy in reducing FCR among early-stage survivors of cancer [63]. CBT was the first approach that was used to build blended interventions in the field of psychological health [64], emerging as an evolution of the classical CBT method and the method that was internet-delivered. Indeed, blended care CBT treatments (BC-CBTs) blend classical therapist-led CBT sessions with internet-delivered CBT components, and it constitutes an integrated therapy, putting together the gains of both approaches while relieving the limits of the single approaches. The interactive presence of the therapist, for instance, allows for the creation of a therapeutic alliance, which in turn is associated with higher motivation to begin and maintain commitment in care [65,66]. Furthermore, the presence of a therapist allows for a fair degree of personalization of treatment plans. Specifically, the therapist using BC-CBT can decide the extent to which face-to-face sessions, live video-based sessions with interactions between therapist and client, and remote digital care tools (lessons and exercises) that can be accessed by the client between sessions can be combined [67,68]. Moreover, internet CBT with the therapist's support leads to better clinical outcomes, more stable results in time, and higher adherence compared with unsupported ones (eHealth) [69,70].

Considering traditional CBT, the Survivors' Worries of Recurrent Disease study is an example of a BC-CBT intervention that combines traditional CBT, which is delivered face-to-face, with web-based activities (or workbook activities) [41,63,71]. This intervention aims to modify the cognitions and behaviors that maintain a high FCR through cognitive reframing, exposure and response prevention, psychoeducation, mindfulness, and relaxation exercises. It includes 5 face-to-face therapy sessions and 3 web-based or telephone sessions with a trained therapist. Participants could access a supportive website or workbook along with the therapy. During the first session, participants conceptualized their personal FCR model, which guided the course of the therapy. Subsequent sessions focused on acceptance, cognitive restructuring, and behavior modification. The final sessions consolidated the progress made by the patient and established a relapse prevention plan. Participants did homework to improve the skills learned during therapy. In an RCT of 88 survivors of cancer with high levels of FCR, participants who received this intervention had significantly less FCR compared with those who were allocated to the standard care group. The effect size was moderate to large (Cohen d=0.76), and both the clinical and self-reported improvements in FCR were higher in the BC-CBT group compared with the control group [63]. Finally, a recent study showed that the Survivors' Worries of Recurrent Disease intervention was effective in reducing FCR compared with treatment as usual in the long term (mean difference −1.787, 95% CI −3.251 to −0.323; P=.02 at 15 months follow-up) [47].

Another example is the blended care for FCR study proposed by Luigjes-Huizer et al [72]. This intervention was developed to be delivered by primary care and not by professionals and institutes specializing in psycho-oncology, as the role of primary care in cancer and survival care is increasing. In particular, general practitioners and mental health workers provided an intervention specifically designed for FCR based on CBT, clinical experience, and input from patients, with web-based modules that focused on normalization, psychoeducation, and self-management. There were 2 modules based on CBT and 5 optional modules (eg, rumination, avoidance, relaxing, reassuring, and undertaking activities). The authors are conducting a 2-armed cluster-randomized trial to evaluate the effectiveness of this intervention [72].

In literature, there is another blended intervention based on traditional CBT, the colorectal cancer distress reduction intervention, which was proposed for survivors of colorectal cancer, which was not specific for FCR [73]. However, we propose this intervention as it comprises 3 separate modules, each of which deals with a specific ailment that the patient may experience, namely distress because of physical consequences, anxiety and FCR, and depression. It aims to facilitate adjustment and coping, reduce distress, and modify cognitions and behaviors. It lasted 14 weeks and comprised 5 individual sessions and 3 telephone consultations, combined with the use of the interactive self-management website, where patients found the homework. Patients discussed their homework completed on the website with a therapist during the individual sessions. In the first session, patients reported their cancer follow-up experiences with a focus on treatment goals, the distress they are experiencing, and their own unmet needs. During the second session, the therapist introduced and explained the basic skills of the CBT applied to the first module. The following sessions included psychoeducation, cognitive restructuring, behavior modification, and relaxation. In the last 2 sessions, the therapist and patient evaluated the reduction of distress and discussed the consolidation of long-term skills. The website included a general introduction module comprising 2 web-based homework sessions focused on introducing CBT and identifying personal goals. A total of 3 specific modules, including different types of self-management activities (eg, psychoeducational scripts, assignments tasks, screening tests, audio clips, and peer videos) and a general closing module focused on goal evaluation and relapse prevention. A 2-arm multicenter RCT of 160 survivors of colorectal cancer with high levels of ailment was initiated by the authors [73]. However, a cognitive behavior therapist decided to treat a 74-year-old male survivor of colorectal cancer with this intervention for 4 months and reported an improvement in psychological distress after intervention [74].

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Regarding blended mindfulness or ACT-based interventions, there have been studies. On the basis of the aforementioned ConquerFear therapy, researchers want to test the efficacy in the short- and long-terms of a therapist-guided version of iConquerFear in reducing FCR and improving QoL in survivors of colorectal cancer. This intervention differs from iConquerFear because of the presence of the therapist through a messenger function with whom patients can communicate asynchronously. The therapist had the role of a motivator and coach, answering the questions and giving feedback on the exercises and written material [75]. A summary of the described studies is presented in Table 3.

Table 3. Blended psychological interventions.

Therapy and authors	Cancer	Study design	Intervention	Results
Traditional CBT[a]				
van de Wal et al [47,63,71]	BC[b], PC[c], or colorectal; survivors; BC, PC, and CRC[d]	Study protocol; RCT[e]; RCT	The SWORD[f] study: 5 individuals 1-hour F2F[g] sessions+three 15-minute web-based sessions based on traditional CBT; UC[h]	• SWORD had a greater effect on FCR[i] than UC with a moderate-to-large effect size (Cohen d=0.76). • SWORD had a greater effect on FCR than UC (mean difference −1.787, 95% CI −3.251 to −0.323; *P*=.02) at the 15-month follow-up.
van de Wal et al [41]	Survivors of BC	Case study	The SWORD study: 7 F2F therapy sessions and 1 telephone session based on traditional CBT	• CBT reduced FCR over time (last follow-up at 12 months after therapy).
Luigjes-Huizer et al [72]	—[j]	Study protocol	BLANKET[k]: 2 CBT modules+5 optional modules; UC	—
Leermakers et al [73]	Survivors of CRC	Study protocol	CORRECT[l]: 5 F2F sessions +3 telephone sessions and an interactive self-management website; UC	—
Döking et al [74]	Survivors of CRC	Case study	CORRECT for 4 months: 5 F2F+3 telephone sessions and an interactive self-management website	• The intervention was successful in reducing the distress of a survivor of cancer.
Contemporary CBT				
Lyhne et al [75]	Survivors of CRC	Study protocol	Therapist-guided iConquerFear: 5 modules; UC	—

[a]CBT: cognitive behavioral therapy.
[b]BC: breast cancer.
[c]PC: prostate cancer.
[d]CRC: colorectal cancer.
[e]RCT: randomized controlled trial.
[f]SWORD: Survivors' Worries of Recurrent Disease.
[g]F2F: face to face.
[h]UC: usual care.
[i]FCR: fear of cancer recurrence.
[j]The type of cancer was not specified.
[k]BLANKET: blended care for fear of cancer recurrence.
[l]CORRECT: colorectal cancer distress reduction.

Discussion

Principal Findings

On the basis of the data presented in the literature, we could infer that a psychological intervention based on CBT is efficacious in managing FCR, especially at posttreatment [4,5,11,12]. Taking into consideration the results of traditional CBT, almost all psychological interventions reduced FCR at postintervention and follow-up regardless of whether they were delivered face to face, on the web, or blended [17,19,41,51,63,71]. Only 1 study did not show any effect on FCR [44]. The remaining studies reduced the level of FCR only at posttreatment [18,20,22-24]. Moreover, there was a lack of data on the efficacy of 2 blended interventions based on traditional CBT [72,73]. To date, no study has simultaneously compared the effectiveness of face-to-face, web-based, and blended interventions.

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Regarding psychological interventions based on mindfulness, only 1 RCT study compared MBCT and eMBCT with usual care and found that MBCT and eMBCT were equally efficacious compared with treatment as usual in reducing FCR [27,76,77]. In general, face-to-face and web-based interventions reduced FCR at posttreatment [28-33,53]. However, there was a lack of studies on blended interventions based on mindfulness.

The retrieved studies in the literature on ACT reported only protocols and feasibility or qualitative studies, with no quantitative data on the effectiveness of such an approach, except for 2 RCTs—1 that provided a face-to-face intervention and the other 1 a web-based intervention [35-37,57]. However, little data on the feasibility of this approach have yielded promising results [56]. Even for ACT, no studies used blended interventions.

Finally, there were 2 programs based on multiple theories: ConquerFear, which clinicians could use face to face, via the web, or as blended; and e-TC. However, there was only quantitative data supporting the efficacy of face-to-face ConquerFear, which had a short and long-time effect on FCR [39,40].

Hence, it was difficult to decide which one to choose. If for interventions based on traditional CBT, there were more data, for the contemporary CBT, the information would still be limited, even more so if we take into consideration the new methods of administering the interventions. Although it is normal given the youth of these new modalities, the need to find effective web-based or blended treatments is increasingly urgent. Furthermore, when it comes to clinical applications, the choice of what should be interactive is not trivial. Indeed, it has been shown that therapists believe that the complexity of patients' problems requires tailored blended treatment. It has also been found that therapists and patients have different points of view regarding what components of the therapy they would prefer to be presented in a web-based-remote way [59]. The parts of treatment that can best be offered on the web or face to face can differ among patients (based on ability, preference, severity, and type of problems) and should thus be considered for each patient individually [59]. For that reason, further studies should explore patients' preferences, not only about the content of the intervention but also the type of intervention (face-to-face, web-based, or blended) they prefer. Another point that is currently lacking in the retrieved literature concerns the stigma of mental health care [78] and how different modalities of delivery of treatments might address this issue. Indeed, mental health stigma has been found to have a small- to moderate-sized negative effect on help-seeking behaviors [79]. Interestingly, web-based interventions might be specifically built to target mental health stigma [80].

Patients and survivors might benefit from blended interventions as they have the potential benefits of face-to-face and web-based approaches. In particular, through a blended intervention, patients might maintain the therapeutic alliance with the therapist without the necessity of meeting them weekly because of, for example, a website in which they have to do some homework to practice the skills learned during the face-to-face session. We believe that an integrative intervention based on both traditional CBT and contemporary CBT would be the best choice.

Future Directions: "Change of Recurrence"

Overview

On the basis of the retrieved evidence, we propose the hypothetical program of an intervention for FCR based on both traditional CBT and contemporary CBT, named Change Of Recurrence, which aimed to improve the management of FCR in patients with cancer and survivors. We would opt for a blended intervention for 3 reasons. First, given the health emergency that we have been experiencing for the COVID-19 outbreak, a blended intervention would allow us to guarantee the safety of the patient who will rarely have to go to the hospital while maintaining the therapeutic alliance, which is fundamental to the effectiveness of any psychological intervention [81]. Web-based interventions are generally well-received by patients with cancer and might enhance comprehensive care [82], whereas blended therapy overcomes the disadvantages of the lack of alliance between eHealth interventions, maintaining the benefits of a face-to-face intervention [59-61] and the cost reduction and increased accessibility of delivering mental health care [83]. Furthermore, in the retrieved literature, the blended modality resulted as effective in reducing the FCR with moderate-to-large effect sizes, maintained across time [41,47,63,71]. Overall, we propose an intervention based on a mix of the key elements that characterize the effective approaches retrieved in the aforementioned literature (CBT, ACT, and mindfulness), delivered through a combination of face-to-face and remote sessions and psychoeducational material.

The program would be structured by first conducting face-to-face sessions at the hospital where the patient is treated or where the survivor undergoes check-ups. This choice could help patients and survivors follow the therapy, especially if we insert the session on the same day that the patient or survivor goes to the hospital for other visits or checks. Indeed, dropout from psychological interventions is a relevant issue to be considered. In addition, we will create a web-based platform comprising 10 modules that can be accessed only when patients finish the previous one. The web-based platform will provide interactive sessions, psychoeducational exercises, and homework. The latter has the objective of trying to render patients as autonomous as possible by applying the techniques learned in the face-to-face sessions.

First Face-to-face Session

During the first face-to-face session, the therapist gets to know the patient, assessing the level of FCR both qualitatively and quantitatively, using the questionnaire Fear of Cancer Recurrence Inventory. In particular, the patient provides details about him or herself and has the opportunity to discuss diagnosis; treatment; recovery; and, in general, his or her experience. Moreover, this will be a psychoeducational session in which the therapist will explain the FCR model, identify the internal and external triggers that increase the FCR, and focus on the patient's FCR experience, particularly on their maladaptive thoughts and coping strategies. At the end of the

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session, the therapist will give the patients a link to the web-based platform. Through the platform, the patients will be asked to write their negative automatic thoughts using a typical 3-column grid.

First Web-Based Session

In the first web-based module, the patient will find a summary of the key concepts addressed during the first face-to-face session and exercises to do, such as writing down the thoughts and actions that he or she performs and that increase the FCR.

Second Face-to-face Session

During the second face-to-face session, the therapist will discuss the patient's homework to clarify the eventual unsolved aspects. In this way, the patient will be encouraged to share his or her experience and start to work on it. The therapist will then introduce the notion of cognitive restructuring, and through the Socratic questioning of cognitive therapy, attempt to challenge the negative automatic thoughts written by the patient during homework. Finally, the therapist and patient will start to reframe negative thinking into alternative thoughts that are more based on reality to explain the homework that the patient will have to do during the week.

Second Web-Based Session

In the second web-based module, the patient will find a summary of the key concepts addressed during the second face-to-face session. The assigned homework that the patient will have to do is to write the negative automatic thoughts using a typical 5-column grid and reframe them into realistic thinking.

Third Web-Based Interactive Session

During the third web-based interactive session, the therapist will review the patient's homework. Then, the therapist and the patient will discuss their thoughts and feelings related to cancer and the actions they will take to get rid of or escape those feelings and those that increase the FCR. In particular, the therapist will provide psychoeducational concepts about worry, explaining the importance of expressing fears. To do that, the patient will have to describe their worst-fear scenario related to cancer, providing thoughts and behaviors that he or she will engage in. Then, the therapist will use metaphors such as the bus metaphor, in which the patient will identify thoughts, feelings, and memories or images as *passengers* persistently challenging cancer. In this way, the patients will learn to actively accept, defuse, and respond compassionately to passengers while not allowing them to dominate their lives. Finally, the therapist will introduce the concept of relaxation, in particular, body scan meditation.

Fourth Web-Based Session

Following the third module, the patient will find a summary of the key concepts addressed during the third face-to-face session on his or her platform. The patient will also be provided with other metaphors and experiential exercises (eg, daily body scan meditation) aimed at improving the strategies built in the web-based interactive session with the therapist.

Fourth Face-to-face Session

During the fourth face-to-face session, the therapist will focus on adaptive coping strategies that enhance acceptance, cognitive defusion, awareness, and psychological flexibility in general. In this way, the patient will begin to become aware and accept thoughts and emotions about cancer, eliminate rigid thoughts and beliefs about cancer, and define personal values and commit to pursuing meaningful activities in line with those values. Finally, the therapist will introduce the concept of mindfulness and its basic principles.

Fifth Web-Based Session

In this web-based module, the patient will be provided with a summary of the key concepts addressed during the fourth face-to-face session, with particular attention to mindfulness. Specifically, the patient will be presented with mindfulness and relaxation exercises through audio and videotape. The audio clips will contain fully automated exercises meant to bring awareness to breathing and bodily sensations. The video clips will provide additional explanations on the techniques, along with some practical guidelines on how to practice during the daytime without clips.

Fifth Face-to-face Session

During the last face-to-face session, the therapist will evaluate the changes that occurred in the patient's emotional and cognitive reactions, making a summary of the current situation and the changes that occurred. The therapist and the patient will talk about the differences between how the patient coped with FCR before the treatment and how he or she copes now. Moreover, the therapist and the patient will draw up an action plan based on the patient's values. Finally, the therapist will build, together with the patient, an exercise schedule to maintain the improvements. The contents of the exercises (audio or video clips) will be available on patients' platforms and will be accessible for a year.

Follow-up Web-Based Interactive Session

A final web-based interactive session will be provided 1 month after the fifth face-to-face session to monitor the psychological state of the patients.

If effective, this program would lead to a time and cost-saving care pathway for treating FCR, putting together the benefits of real-time interaction with the clinical staff and the ease of having tailored clinical materials available daily to allow for a continuous improvement.

Conclusions

To conclude, this overview has some limitations. Regarding the first part (review of the psychological interventions), the limitations of the methodology of the included studies and between-study heterogeneity reduced the overall strength of the evidence. Moreover, some of the studies were selected from other systematic reviews, whereas others were selected manually. Regarding the second part, it is only a preprotocol that must be evaluated by experts and patients.

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Conflicts of Interest

None declared.

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Abbreviations

ACT: acceptance and commitment therapy
AIM-FBCR: Attention and Interpretation Modification for Fear of Breast Cancer Recurrence
BC: breast cancer
BC-CBT: blended care cognitive behavioral therapy treatment
CBT: cognitive behavioral therapy
eMBCT: internet-delivered mindfulness-based cognitive therapy
FCR: fear of cancer recurrence
MBCT: mindfulness-based cognitive therapy
MBSR: mindfulness-based stress reduction
QoL: quality of life
RCT: randomized controlled trial

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Original Paper

Understanding the Information Needs of Patients With Ovarian Cancer Regarding Genetic Testing to Inform Intervention Design: Interview Study

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Abstract

Background: Experts in gynecological cancer care recommend that all patients with invasive or high-grade ovarian cancer (OC) undergo genetic testing. However, even patients who intend to take or have taken genetic tests have many unaddressed information needs regarding genetic testing. Existing genetic counseling falls short of adequately addressing this challenge.

Objective: This study aims to investigate the genetic testing–related information needs of patients with OC to inform the design of interactive technology-based interventions that can enhance communication of genetic testing information to patients.

Methods: We interviewed 20 patients with OC who had taken genetic tests and gathered genetic testing–related messages from an active OC web-based community. The interview transcripts and web-based community messages were analyzed using the qualitative content analysis method.

Results: Data analyses produced a comprehensive taxonomy of the genetic testing–related information needs of patients with OC, which included five major topic clusters: knowledge of genetic testing as a medical test, genetic testing process, genetic testing implications for patients, implications for family members, and medical terminology. Findings indicated that patients wanted to receive information that was relevant, understandable, concise, usable, appropriate, sympathetic, and available when needed. They also preferred various channels to receive information, including internet-based technologies, print, and conversations with health care providers.

Conclusions: Patients with OC need a range of information to address the uncertainties and challenges that they encounter while taking genetic tests. Their preferences for channels to receive information vary widely. A multichannel information delivery solution that combines both provider-led and peer-to-peer education models is needed to supplement existing genetic counseling to effectively meet the genetic testing–related information needs of patients with OC.

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KEYWORDS

patient information needs; consumer health informatics; ovarian cancer; genetic testing; genetic counseling; mobile phone

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Introduction

Ovarian cancer (OC) is the second most common gynecological cancer in the United States [1]. Nearly 25% of OC cases are due to hereditary cancer syndrome as a result of breast cancer gene mutations (*BRCA1* and *BRCA2*) and Lynch syndrome [2,3]. The National Comprehensive Cancer Network and the Society of Gynecologic Oncology recommend that all patients with invasive or high-grade OC undergo genetic testing [4,5] as knowledge of gene mutations can inform targeted treatment [6] as well as cancer screening and prevention options for at-risk family members [7].

Nevertheless, the genetic test uptake rate among patients with OC falls short of expectations. For example, 2 studies reported that only 15% to 20% of all women diagnosed with OC underwent genetic testing for *BRCA1* and *BRCA2* [8,9]. Another more recent estimate of the testing rate among newly diagnosed patients with breast cancer and OC was 53% [10]. Although attention needs to be placed on promoting genetic testing uptake among patients with OC and their family members, there are unmet information needs among those who intend to take or have taken genetic tests that also need be addressed. For example, studies have reported that some patients with OC have never heard of *BRCA1* and *BRCA2*, are unaware of the relevance of genetic testing for themselves and their families, or underestimate the actual risk of a hereditary link to their diagnoses [6,11,12]. Studies have also found that some patients with cancer and patients at risk for cancer had concerns about genetic testing–associated risks, such as insurance discrimination, privacy infringement, and emotional distress [11,13,14].

Communication of information concerning cancer genetics and genetic services to patients needs to be improved to address patients' literacy gaps and risk concerns to enhance patient satisfaction and sense of empowerment. Some interventions have been conducted [15-19]; however, most have focused on exploring noninferior alternative genetic counseling delivery models (eg, group counseling) to the traditional one-on-one face-to-face model, paying little attention to the materials delivered. Analyses of genetic counseling sessions have revealed that genetic counseling communication is largely provider-driven, centering on providing biomedical information and failing to consider patients' information, communication, and psychosocial needs [15,20-27]. Furthermore, most interventions were delivered through traditional information channels (eg, booklets and telephone) or basic interactive technologies (eg, videos) [19], missing the potential that interactive web-based technologies can offer. Thus, there is significant room for designing web-based interventions to address patients' genetic testing–related knowledge gaps and concerns.

Designing effective technology-based interventions requires a thorough understanding of patient information needs [28,29]. We define patients' information needs regarding genetic testing as knowledge gaps that patients perceive or experience as preventing them from accomplishing genetic testing–related activities or goals. These knowledge gaps may result from cognitive and affective uncertainties and may be a result of environmental (including institutional, cultural, and societal) constraints [30-33]. Information quality (IQ), defined as "users' perception of the quality of information presented on a Web site" [34], has been identified as a significant information-related factor that precedes the formation of people's trust in and intention to use information systems [35-37]. The fulfillment of information needs is not possible if IQ is low. Thus, we also explore patients' expectations of the quality of genetic testing–related information. In addition, we explore patients' preferences concerning information delivery to fulfill our aim to inform system design. The specific research questions are: (1) Which topics of information do patients with OC need to be informed about regarding genetic testing? (2) How do patients characterize their preferences for the quality of genetic testing–related information? (3) From which information channels, media, or platforms do patients prefer to receive genetic testing–related information?

Methods

Owing to limited research on this subject, we adopted a qualitative research design consisting of two methods: interviews and analyses of web-based community posts.

Interviews

Participant Recruitment

The participants were women who had been diagnosed with OC and had undergone genetic testing. Recruitment was performed in 3 ways. The first was a chart review by a clinical research assistant at the Dell Medical School at the University of Texas (UT). More than 30 eligible patients who received treatment from a physician in the LIVESTRONG Cancer Institutes at the school were contacted. Reasons for not participating included a lack of interest or energy, language barriers (non–English-speaking), and a lack of resources (car, computer, or webcam). Second, we posted email recruitment messages to the mailing list of the National Ovarian Cancer Coalition Austin and San Antonio Chapter. Third, we adopted word-of-mouth and snowballing recruitment strategies. Recruitment efforts using all 3 venues spanned the entire research process (data collection and analysis) and halted when a theoretical saturation of the data was observed. The data were deemed saturated when no new genetic testing–related information needs, IQ, or information delivery themes emerged from the data. A total of 20 patients with OC participated in the interviews, of which 8 (40%) were recruited through the chart review, 10 (50%) were recruited through the mailing list, and 2 (10%) were recruited through word-of-mouth.

Interview Design

The interview protocol had three components: a demographic questionnaire, a semistructured interview, and a co-design session. The guide for the interview and the co-design session is provided in Multimedia Appendix 1. The demographic questionnaire collected the participants' background information, including demographics (eg, age, race, ethnicity, and education), cancer diagnoses, and genetic test results. In the semistructured interviews, the participants recalled their

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genetic testing process (from when they were prescribed the test to receiving the test results) and experience (including motivations, emotions, interactions with health care providers and family and friends, and challenges). They were also asked to describe their genetic testing–related information behaviors, including information needs, information sources, and information-seeking efforts.

In the co-design session, the participants reviewed and commented on a mockup website that offered genetic testing–related information while imagining that they were co-designing the website for patients such as themselves. They were also asked to describe any additional content that they thought should be included on the website, their expectations of IQ, and how they wanted genetic testing–related information to be presented and delivered to them. The co-design session was used because people sometimes experience difficulty in perceiving and articulating their information needs and preferences [38]—interactions with information sources may make some information needs and preferences for IQ and information delivery more visible [39]. Questions concerning IQ were framed based on a successful validation of the information system success model by DeLone and McLean [29], which identifies six IQ dimensions: availability, usability, understandability, relevance, format, and conciseness [40,41]. The initial mockup was paper-based, created based on a review of studies on the genetic testing–related information needs of patients with OC (Figure 1). The paper mockup was later developed into a digital mockup (Figure 2) based on ongoing analyses of the interviews. The content displayed on the mockups was drawn from trustworthy sources such as the National Cancer Institute and the Centers for Disease Control and Prevention.

Figure 1. A sample page of the paper mockup. BRCA: breast cancer gene; MLH: mutL homolog.

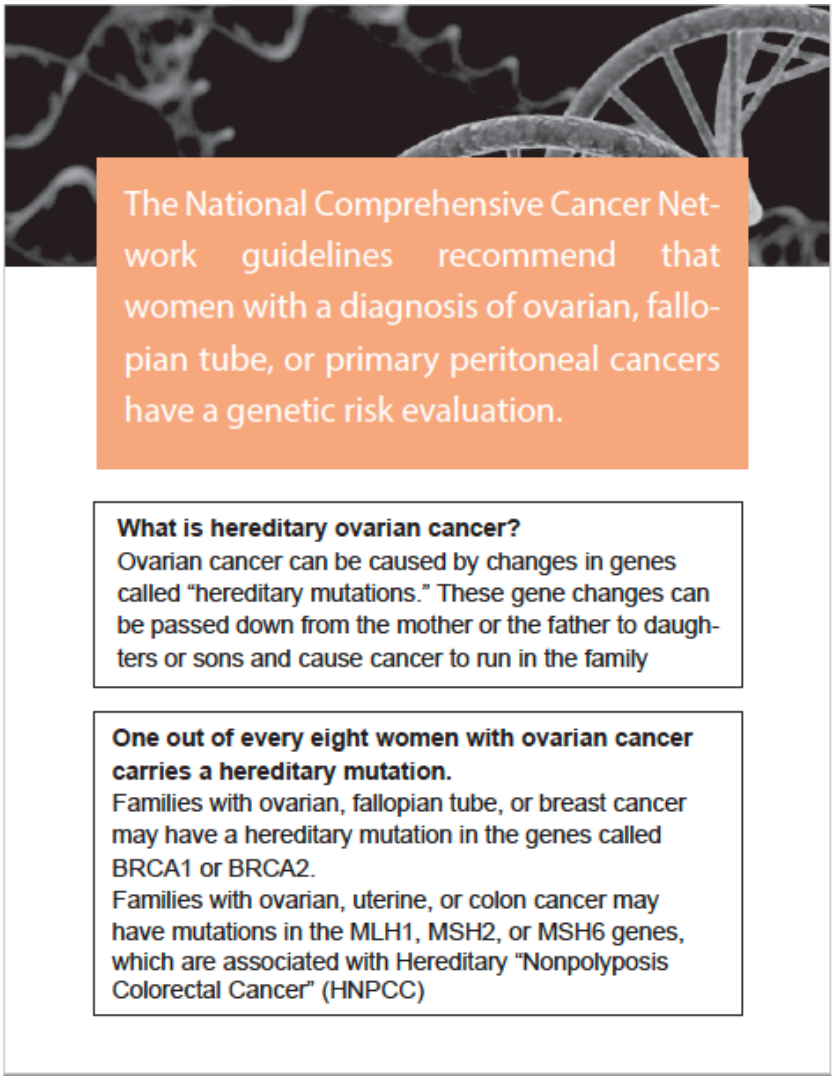

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Figure 2. A sample webpage of the digital mockup website.

The Interview Process

The interviews were conducted between February 2019 and October 2020. The first 6 participants (6/20, 30%) were interviewed in 5 face-to-face focus groups that took place in a private conference room at the UT campus. Each focus group consisted of 2 participants. A total of 2 participants (2/6, 33%) took part in 3 focus groups as we were not able to complete both the interviews and the co-design activities in 1 session. The other 4 patients (4/6, 67%) participated in 1 focus group session each. Upon arrival, researchers greeted the participants, gave them an introduction to the project, and asked them to review the consent form. The participants were encouraged to ask clarifying questions when needed. After providing consent, the participants completed the demographic questionnaire. They were then interviewed about their genetic testing process and experience as well as genetic testing–related information-seeking activities. The focus group interview format was adopted as it allows for interactions between participants with the goal of helping participants recall and elaborate on their genetic testing experience. The interviews were followed by the co-design session. Upon completion of the co-design session, the participants received a US $30 Amazon gift card.

Owing to the COVID-19 pandemic, the subsequent 13 interviews (13/20, 65%) were conducted one-on-one through the Zoom web conferencing platform, and 1 participant (1/20, 5%) was interviewed through emails. In these interviews, the participants completed the consent process and the background questionnaire on the web on Qualtrics (Qualtrics International Inc) before the interview. The Zoom interviews followed the same procedure as the focus groups. For the email interview, we sent the questions to the participant, and she responded with written answers. In total, 2 researchers (YZ and SY) reviewed and discussed the answers and then asked clarifying questions by commenting on the answers. She then replied to the clarifying

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questions. A total of 3 rounds of email correspondence took place. The URL of the digital mockup website and the questions that we asked in the co-design session were then emailed to the participant. She answered those questions. Similarly, we asked clarifying questions by replying to her answers.

The focus group and Zoom interviews lasted 40 minutes to 2 hours. Each interview was conducted by at least two researchers, audio-recorded, and later transcribed. The researchers held a 20- to 30-minute debriefing session after each interview to generate main themes related to the research questions and insights to inform the design of the digital mockup website.

Web-Based Forum Message Analysis

Social media platforms (eg, web-based health forums and social question and answer platforms) are sources for collecting authentic consumer health information needs [42]. Web-based posts are also considered an ecologically valid means of eliciting user needs for technological design [43]. We searched the OC community on the American Cancer Society's Cancer Survivor Network (CSN) to identify genetic testing–related posts. The keywords used for the search included *genetic testing*, *genetic counseling*, *BRCA*, and *DNA testing*. The search identified 210 messages. We manually collected these messages, read them, and, of the 210 messages, we retained 25 (11.9%) that contained patients' genetic testing–related information needs for analysis. These messages were posted by 25 unique IDs between December 2008 and June 2018. Excluded posts included answers to the questions posted, genetic testing resources, the patients' own OC experiences, and family members' concerns for themselves.

Data Analyses

The interview transcripts and CSN messages were analyzed using both inductive and deductive approaches to the qualitative content analysis method [43]. First, we imported the interview transcripts and web-based forum messages to MAXQDA 2018 (VITERBI Software GmbH), a qualitative data analysis software. Initially, YZ coded 5 interview transcripts deductively by following the definition of information needs (ie, knowledge gaps that patients perceive or experience as preventing them from accomplishing genetic testing–related activities or goals) and the IQ dimensions outlined in the study by Petter et al [41] (including availability, usability, understandability, relevance, format, and conciseness). An inductive approach was then applied to generate subcategories of genetic testing–related information needs, additional categories of IQ dimensions, and technological platforms for information delivery [44,45]. A codebook was developed to keep track of and explicate the coding system.

CT and SY applied the codebook to independently code 2 interview transcripts. The research team then held several collaborative coding sessions to discuss codes, paying special attention to reconciling codes to reduce overlap and redundancy between subcategories [45]. This effort resulted in a revised codebook. SY, CB, and YZ each revisited the codes that they had assigned to the transcripts by applying the new codebook. Each researcher then coded a subset of the remaining transcripts. SY coded the forum posts. All codes were validated by a different coder to enhance coding reliability, and disagreements were resolved based on discussions between all research team members.

Ethics Approval

The University of Texas at Austin Institutional Review Board approved the study.

Results

Interview Participants

Most participants were aged >40 years (19/20, 95%), White (16/20, 80%), non-Hispanic (13/20, 65%), and had a college or postgraduate degree (13/20, 65%; Table 1). Their cancer stage at diagnosis varied. Of the 20 participants, all of them (100%) had previously undergone germline genetic testing, and 6 (30%) had also undergone somatic genetic testing. Most of these tests (13/20, 65%) were conducted in the past 3 years (2018-2020). The test results varied.

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Table 1. Participant characteristics (N=20).

Characteristics	Participants, n (%)
Age (years)	
<40	1 (5)
40-49	6 (30)
50-59	5 (25)
60-69	6 (30)
70-79	2 (10)
Race or identity	
White	16 (80)
American Indian or Alaska Native	1 (5)
Mexican-American	1 (5)
Not reported	2 (10)
Ethnicity	
Hispanic or Latino	7 (35)
Non-Hispanic or Latino	13 (65)
Level of education	
<8 years	1 (5)
8-11 years	1 (5)
Post–high-school training other than college	2 (10)
Some college	3 (15)
College graduate	7 (35)
Postgraduate	6 (30)
Cancer stage when diagnosed	
1	3 (15)
2	3 (15)
3	6 (30)
4	5 (25)
Not reported	3 (15)
Year in which the most recent genetic test was taken[a]	
2018-2020	13 (65)
2015-2017	5 (25)
2012-2014	2 (10)
Test results	
Germline positive	6 (30)
Germline negative	7 (35)
Variants of uncertain significance	1 (5)
Germline negative and somatic positive	1 (5)
Germline and somatic negative	5 (25)

[a]All 20 participants had taken a germline test, and 6 (30%) had also taken a somatic test.

Information Need Topics

Patients' genetic testing–related information needs coalesced around five topic clusters: basic knowledge of genetic testing as a medical test, genetic testing process, implications of genetic testing for patients, implications for family members, and medical terminology.

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Basic Knowledge of Genetic Testing as a Medical Test

The cluster of basic knowledge of genetic testing included two topic categories: basic features of genetic testing and standards and regulations (Table 2). Example questions were extracted from the data and rephrased for conciseness and clarity.

Regarding genetic testing features, the patients wanted to know what genetic testing is, what it does, and its benefits and potential risks. In terms of standards and regulations, the patients wanted to learn who approves genetic testing, who is qualified to provide it and the providers' qualifications, and relevant government standards and regulations.

Table 2. Patient information needs regarding basic knowledge of genetic testing (GT) as a medical test.

Category and subcategory	Example questions
Basic features of GT	
What is GT?	• How are clinical forms of GT different from direct-to-consumer GT? (I[a]) • What are the distinctions between the different methods of GT recommended for cancer patients? (C[b]) • What are the current tests for ovarian cancer? (I)
What does GT do (ie, functions)?	• What does GT do? (C) • What does GT uncover or look for? (I) • Can GT determine or test for cancer? (I) • Can GT determine other diseases? (I) • How is the information from GT used? (I)
Benefits (why GT?)	• Why would you want to be genetically tested? (I) • Why is GT important? (I) • How is GT beneficial in saving lives and helping families get pre-screening to detect cancer sources? (I) • What benefits can GT results bring to the treatment of OC[c]? (I, C)
Risks	• What are the possible risks of GT? (I)
Standards and regulations	• Who approves GT? Is the FDA[d] involved? (I) • Who is qualified to offer this service? What are their qualifications? (I) • What are the standards and regulations related to GT? (I)

[a]Indicates that the example is from the interviews and co-design sessions.

[b]Indicates that the example is from the web-based community message analysis.

[c]OC: ovarian cancer.

[d]FDA: Food and Drug Administration.

Genetic Testing Process

This topic cluster included three topic categories: financial demands, taking the test, and obtaining results (Table 3).

Cost was often the patients' first concern when considering taking genetic tests. As an interview participant put it, "cost was my first question." They wanted to know whether their insurance covered the test and, if not, how much they must pay. In several cases, the participants did not have insurance, and third parties (eg, foundations) subsidized the cost. A few participants mentioned that they had considered not taking genetic tests if the cost was not covered by insurance or third parties.

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Table 3. Information needs regarding the genetic testing (GT) process.

Category and subcategory	Example questions or comments
Financial demands	
Cost	• What is the cost of GT? I would want to know the costs right up front. (I[a]) • Do I have to pay for GT? (I) • The hard thing I've noticed in the US is that they often can't tell you even how much your cost is, because it depends on your insurance and all these ridiculous things. (I)
Insurance coverage	• I would want to know whether my insurance covers GT. (C[b], I) • Does Medicare cover GT? (C)
Taking a GT	
Who does GT?	• Can a regular doctor perform GT for cancer genes? (C) • Who is doing the test? Who are they? (I) • Who are the testing companies? Can we choose which one to use? (I) • What lab will you need to go to take the test? (I)
Procedure and test details	• I would be interested in knowing how GT is done and have a better understanding of that. (I) • What exactly happens in the lab? (I) • How much blood will be drawn? Is there an alternative to a blood draw? (I) • Is it painful? (I)
Obtaining results	
Receiving GT results	• What is the timeframe for getting GT results? (I) • Will I be contacted when they find new information from the test results? If so, how, and when will I be contacted? (I) • Can I get a copy of the GT results? (I)
Genetic counseling	• How long do I have to wait to see the genetic counselor? (C) • What questions should I ask during a genetic counseling session? (C) • I'm not sure whether or not to have my GT results interpreted. (C) • Who would I talk to about GT to understand if my ovarian cancer was genetic or not? (C) • Who will interpret the results for me? (I) • What is the significance of a particular result, like VUS[c]? (I)

[a]Indicates that the example is from the interviews and co-design sessions.

[b]Indicates that the example is from the web-based community message analysis.

[c]VUS: variants of uncertain significance.

Regarding taking the test, the patients wanted to know who recommends and orders genetic testing, who conducts genetic testing, the testing companies involved, and the laboratories that perform the test. They also wanted information about test procedures, including how it is done, whether a blood draw is needed, and whether it is painful.

Information about when and how they receive the test results and whether they can obtain a copy of the results was also needed. Some patients knew about genetic counseling and asked specifically about it, including when to receive genetic counseling and what questions to ask. Some patients hesitated to pursue genetic counseling and sought peers' opinions (through web-based communities). Some patients were not aware of genetic counseling and wondered who could help interpret their genetic test results.

Implications of Genetic Testing for Patients

Overview

This topic cluster included five topic categories: cancer causes, clinical implications, genetic discrimination, lifestyle, and communication with family (Table 4).

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Table 4. Information needs concerning the implications of genetic testing (GT) for patients.

Category and subcategory	Example questions or comments
Cancer causes	• Do I have a genetic mutation? Am I a carrier? (I[a]) • I was curious to see if I had a genetic mutation for the cancer to begin with. (I) • What caused my cancer? Genetic mutation or my diet? (I) • My GT result indicates that I am at risk for breast cancer, but I had ovarian cancer, not breast cancer, I need an explanation. (I)
Clinical implications	
Treatment	• Can GT results affect my cancer treatment? If so, what are the effects? (I, C[b]) • What type of chemotherapy do you get if positive for a BRCA[c] mutation? (C) • Will I have a harder time fighting off the cancer given that I have tested positive for the BRCA2 mutation? (C)
Preventative strategies to reduce cancer risks	• What preventative measures can be done if the results come out positive? (I, C) • How do I know if I should follow the doctor's advice regarding preventative surgery? (C)
Genetic discrimination	
Insurance discrimination	• Does anyone know of cases of insurance companies using a GT result to deny benefits to subscribers? (I) • Could the GT result be used against me to deny my healthcare or life insurance coverage? (I, C) • Is this going to affect my insurance later in my life? Am I going to have to pay more money somehow? (I) • Who has access to my GT information? Are there laws to protect us from genetic discrimination [vis-à-vis health insurance]? (I, C)
Employment discrimination	• Can my GT result records be used to deny my employment? (I)
Lifestyle	• Is there anything I can do in relation to lifestyle and diet to minimize any problems that might rise from the GT being positive for a mutation? (I) • If my genetic testing is abnormal, are there lifestyle or diet modifications that are helpful to reduce the risk of developing cancer? (I)
Communication with family	• I was worried like if I had genetic mutations, at what point do I discuss this information with my children? (I) • How do I approach my family and talk to them about GT results? (I, C)

[a]Indicates that the example is from the interviews and co-design sessions.

[b]Indicates that the example is from the web-based community message analysis.

[c]BRCA: breast cancer gene.

Cancer Causes

The patients showed a great deal of interest in seeking answers to the following question—*what has caused my cancer*—in light of their genetic test results. When they had mutations related to breast cancer but not OC, they wanted explanations for why they had developed OC. When the genetic test results were negative, some patients questioned whether it was their lifestyle (eg, diet) that caused the cancer.

Clinical Implications

Questions concerning clinical implications mainly focused on 2 aspects. The first was how the results can inform treatment. Questions ranged from general inquiries about whether test results would affect the treatment to questions about specific therapies. For example, a patient posted the following on the CSN community:

> *Within the last couple of days there was new information about BRCA women who had ovarian cancer (I think BRCA2 not sure) and new chemotherapy available for that. Has anyone else who has ovarian cancer gone for BRCA testing? If so, what type of chemo did you get?*

The second aspect was what preventative measures could be taken to reduce the risk of other cancers, mostly breast cancer. Questions ranged from general inquiries about what preventative measures are available to more specific inquiries about preventative surgeries (eg, prophylactic mastectomy). The following message from the CSN community is an example:

> *[Has] anyone had to undergo a prophylactic mastectomy to PREVENT breast cancer? I have tested positive on genetic testing after stage 3 ovarian cancer and now [doctors are recommending] the mastectomy. Have many questions!*

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Genetic Discrimination

Patients were concerned about who has access to their data, whether the data could be used to deny them health or life insurance or raise insurance costs, and whether there are laws to protect them from such discrimination. Worries about potential employment discrimination were also expressed.

Lifestyle

In relation to lifestyle, patients expressed a need to know how they can modify their lifestyle (eg, diet) to minimize risks incurred by genetic mutations and to manage treatments.

Communication With Family

As a patient's post on the CSN community illustrated, "it is a horrible thing to have to tell your family members they [too] might [develop cancer]." Some patients expressed a need to gain knowledge about how and when to talk with family members about their genetic test results, particularly if the results were positive for a mutation.

Implications of Genetic Testing for Family Members

Overview

Patients' information needs regarding genetic testing implications for family members focused on family members' cancer risks and on concerns regarding insurance discrimination and emotional distress (Table 5).

Table 5. Information needs regarding the implications of genetic testing (GT) for family members.

Category and subcategory	Example questions or comments
Cancer risks	
GT screening	• Who (which family members) should be tested? (I[a], C[b])
Prevention and monitoring strategies	• What course of action can be taken [for family members] if I tested positive? (I, C)
Insurance discrimination	• Will my family members be denied insurance? (C) • How would positive results affect my children when they need their own healthcare? (I)
Emotional distress	• Will my family be living in fear as a result of positive GT results? (C) • I worry that my family will be living in fear. (C)

[a]Indicates that the example is from the interviews and co-design sessions.

[b]Indicates that the example is from the web-based community message analysis.

Cancer Risks

Information concerning cancer screening for family members was a category of information needed. For example, an interview participant noted the following:

> *When my test returned as positive. I have only one concern. I worry about passing [the genes] to my kids.*

She later added that "the next [question] is who should be tested?"

Some patients also wanted to be informed of cancer prevention and monitoring strategies (eg, surgeries) that family members can follow if their genetic test results are positive. For example, an interview participant said the following:

> *[My niece] had 3 children and she's done having children. Her genetic makeup is kind of similar to ours, and probably that would be something she could have monitored easily and if she did carry that and was concerned, she could have her ovaries removed before she had any problem. I think if you find you are predisposed of having breast cancer, there are somethings you can do to minimize your risk. My sister is correct that knowledge is power.*

Insurance Discrimination

Some patients worried about insurance discrimination against their family members. The following post on the CSN community demonstrates this concern:

> *I had the genetic testing in March and some of my family members were [leery] of* [being denied insurance benefits]

Emotional Distress

Some patients worried that their genetic test results may cause stress to their family members. For example, a patient commented the following on the CSN community:

> *I have a lot of cousins, and none have gotten cancer even though most of us are in our 50s. I certainly would hate to think of my 2 daughters (ages 14 and 22) having to suffer from cancer. I wouldn't want them to feel afraid of that. So, it is unlikely I would do any sort of genetic testing.*

Terminology

The need to understand genetic testing–related medical terminology cuts across different stages of the genetic testing process. A CSN community user mentioned the difficulty of articulating requests for genetic testing:

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> *I want to call my doctor to give me a written request for [cancer] genetic testing. What should I ask for? Can't seem to find the exact terminology on the [Internet] and I want to be sure it's correct.*

An interview participant called such terms "the big words" and mentioned difficulties in understanding genetic test results:

> *I looked up [online] some of the words [in my GT results] to see what they mean. I don't know any of them.*

Patients' Preferences Concerning IQ

The participants expressed preferences for seven IQ dimensions: relevance, understandability, conciseness, usability, appropriateness, being sympathetic, and availability (Table 6).

Table 6. Patient preferences concerning information quality.

Information quality dimension	Example participant comments
Relevance	• "[A website is of interest to me when it is about] BRCA[a] [and] linked to ovarian cancer." [Participant 18]
Understandability	• "The basics are good. The nurses break it down to basics and to my level." [Participant 2] • "I wish they would have just broken it down in layman terms for middle aged women that aren't so tech savvy. Just simple, simple words." [Participant 18]
Conciseness	• "I think your text is informative, but not overwhelmingly long. It's short and concise and to the point." [Participant 14] • "People may be fearful to look at something that's a little more detailed." [Participant 17]
Usability	
Tables, bullets, and white spaces	• "Maybe a table would help. Genetic drives of cancer. There is a lot of good statistics in there...I know I tend to look at tables and statistics." [Participant 17] • "I like it because it's nice and clean and has a lot of white space and bullets." [Participant 9] • "I really liked that you have a lot of white space, you know, on the page because I think that that helps make it less intimidating." [Participant 18] • "There's a fair amount of space. I mean, it's not overloaded." [Participant 15]
Additional sources	• "I think...providing basic information and with links to find out more. Someone wants to kind of expand on that basic information." [Participant 2] • "...have the ability to go deep or stay high." [Participant 16]
Appropriateness	• A comment on an image used on the mockup webpage about test results: "She looks very happy for having such a serious conversation. She just looks just a little too happy for that. I mean, it, as I remember, it was, it was stressful, not horrifically stressful, but it was stressful waiting for the results." [Participant 16] • "I don't know if I'd want to show [a picture that shows tubes containing blood] just because of those few people I've met that are so fearful of blood." [Participant 13]
Being sympathetic	• "...what would get my attention would be if there was something that said, Hey, you don't have to have cancer [to get genetic testing]. Don't be afraid of this. It's not a death sentence. It's not, you know, you're looking into a crystal ball or having someone read your future." [Participant 18]
Availability	• "[The mockup website] probably would have been a comfort to me to be able to go and look these things up. And just because so many times in the beginning, I found myself going back over the same stuff over and over, what does this mean? What does this mean? And I think, well, I already read that, but did I miss something when I read it." [Participant 8] • "It's something that, you know, that you can take with you, especially cause when you're, you're going somewhere and all of a sudden you have a question about, well, was that really what I thought it was and you can go back and look at it." [Participant 17]

[a]BRCA: breast cancer gene.

Relevance refers to the information provided being directly relevant to OC-related genetics and genetic testing. *Understandability* refers to whether the information is easy to understand. The participants used terms including "basic," "simple," "self-exploratory," "straightforward," "layman's terms," and "easy to digest" to express this expectation. *Conciseness* indicates that the information should be brief and succinct. Too much detail may incur a sense of information overload and discourage some patients from reading further.

Usability indicates that the information should be user-friendly. In this study, the concept was mostly related to the information presentation format. The participants preferred structured formats—tables, bulleted lists, and white spaces—as they made the text less "intimidating" and were easier to follow. The participants weighed usability over the amount of information they could receive. They suggested the use of hyperlinks to expand beyond basic information when needed.

Appropriateness was mainly about the images used in this study. The participants expressed concerns about several images on

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the mockup website, commenting that they instilled fear or were inappropriate for cancer contexts (eg, one image showed tubes containing blood and the other image showed a character with a seemingly happy smile that was perceived to be unfit for a medical consultation setting). Being *sympathetic* suggested that the participants wished that the information had an understanding and encouraging tone, showing consideration of information seekers' emotional states (eg, fear and need for hope). *Availability* represented the participants' expectations that the information source would be available for them to access whenever and wherever needed.

Patients' Preferences Concerning Information Delivery

Table 7 shows the participants' preferences for channels from which to receive genetic testing–related information.

Table 7. Patient preferences concerning information delivery.

Channel and subcategory	Specifics
Digital technologies	
Media and platforms	• Internet • Websites • Email • Mobile apps • Patient portals • Social media
Devices	• Computers (laptop or desktop) • Smartphones • Tablets
Paper-based prints	• Pamphlet or brochure • Written information to take home
Health care providers	• Gynecologist • Oncologist • Nurse navigators • Nurses • Genetic counselors • Insurance company • Genetic testing company

Some participants preferred to receive information from digital technologies, varying from the internet (in general), websites, and email to patient portals, social media, and mobile apps. Some participants valued the social interaction affordances offered by certain digital technologies. For example, participant 12 suggested the following:

> *Probably on a website, even on an app. I mean, because you know, it wasn't until I was diagnosed with cancer that I realized there's so many apps out there that talk to other people going through what you're going through...And they post like what they're going through, what kind of meds they're on, what kind of chemo they took. And it kind of makes you understand what other people are going through. And so, it kind of helps you, and then you know if there was something like that too [about GT], and that would help person.*

Their preferred devices for accessing information also differed and included laptop or desktop computers, smartphones, and tablets. The participants saw a need to make the information source adaptive to these different screen sizes, as participant 7 suggested:

> *Just make sure it is mobile friendly as you don't know if folks will access it via a desktop computer, laptop, tablet or their smart phone.*

Some participants preferred to receive pamphlets, brochures, or some other form of written information to take home. For example, participant 16 indicated the following:

> *I would want it printed. Okay. I'm still old school...in spite of designing computer systems for a living. I still like paper.*

Participant 17 commented the following:

> *I think something that you can save I think written is good.*

Other participants preferred to receive genetic testing–related information directly from health care providers, including gynecologists, oncologists, nurse navigators, nurses, genetic counselors, insurance companies, or genetic testing companies. For this channel, the preference was for information to be conveyed through face-to-face meetings, phone calls, or written materials such as pamphlets. For example, participant 12 described the following:

> *Well, I've been seen at a gynecologist since I was. I think the very first time I went to go see a gynecologist, I was maybe like 23 or 24. And I had never heard of genetic testing until when I got diagnosed with the cancer. So, I think somewhere in between, you should be told, you know, Hey, get this, you know, you might help me. You know, I, cuz I know like, like when my niece was in her teens, they were*

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> *offering that shot for the cervical cancer. I don't remember what it's called. Yes. It wasn't there when we were, when we were growing up, it's something fairly new. And I think that would probably have helped many people along the way, you know? So, anything that could prevent something like this, I think is good.*

Participant 17 described that she expected to receive genetic testing–related information from a nurse or a staff member in oncologists' offices:

> *I think someone separate would actually be better because I think that really, and truly the doctors are trying so hard to save your life, that you get super focused in on that. And I think, I think someone like maybe a nurse maybe just a certain staff member at the doctor's office.*

Discussion

Principal Findings

This study investigated the information needs of patients with OC related to genetic testing and their preferences for IQ and information delivery to inform interventions to enhance the genetic testing experience and sense of empowerment of patients with OC. This makes 3 major contributions to the literature, as detailed below.

Taxonomy of the Genetic Testing–Related Information Needs of Patients With OC

Previous studies on the genetic testing–related information needs of patients with OC are limited. They have mostly used the survey method [11,46,47] and focused on genetic counseling instead of the patients' entire genetic testing process [12], limiting the range of information needs identified. We explored patients' information needs throughout their genetic testing process, from when they were prescribed the test to when they received and reflected on the test results, using multiple qualitative methods, including interviews, participatory co-design activities, and the analysis of genetic testing–related messages on an active OC web-based community. Together, these methods afford in-depth inquiry of the information needs of patients with OC, leading to a comprehensive taxonomy of their genetic testing–related information needs. This taxonomy confirmed many genetic testing–related information needs of patients with breast cancer and OC reported in previous studies, such as the purpose of testing, implications for treatment decisions, treatment options, time frame for results, and the availability of predictive testing for relatives [11,12,14,15,46-49]. It also revealed numerous topics that have been less reported in the literature, such as genetic testing–related standards and regulations, financial demands, medical professionals involved in genetic testing, communication with family members about genetic testing, and the impact of genetic test results on patients' lifestyle [15].

Many of the needs identified in the taxonomy are consistent with expert genetics and cancer health professionals, who agree that information about inheritance, cancer risks, and management are key messages for patients with cancer [50]. Clinical guidelines for genetic counseling also recognize that some topics in the taxonomy should be covered in pre- and posttest genetic counseling, such as psychological issues, including coping with disclosure of test results, and social issues, including the impact of testing on insurance, employment, and family relationships [51]. Nevertheless, it is still important to recognize patients' perspectives and priorities regarding their own information needs, considering that patients continue to report various unmet needs years after the release of clinical guidelines for genetic counseling [11,12,14]. Thus, this taxonomy can serve as a patient-centered road map for creating information architectures for interventions that address the information needs of patients with OC.

It is also important to acknowledge the limitations of the taxonomy in light of the methods we adopted. The semistructured interview and participatory co-design methods afford the ability to delve deeply into a set of issues, probe and ask follow-up questions, and connect ideas in real time as a discussion unfolds; however, the methods assess information needs retrospectively, increasing the chance that the participants might not have recalled all the information needs that came up before, during, and after the genetic testing process. The web-based community message analysis can help compensate for the limitations of the interviews as the messages represent patients' real-time information needs; however, the number of posts that we were able to collect was constrained because of a lack of discussion on this topic among users of the chosen web-based community.

IQ as an Attribute of Information Needs

Guided by the information system success model by DeLone and McLean [29], we identified seven IQ dimensions that patients with OC deemed important: relevance, understandability, conciseness, usability, appropriateness, being sympathetic, and availability. This finding is consistent with the finding of previous empirical studies that patients with cancer prefer brief, straightforward, personalized, and positive information for genetic testing communication [6,47]. Nevertheless, we examined patients' IQ preferences from a more systematic approach (ie, both theory- and data-driven). These IQ dimensions together offer insights on how information should be written, organized, and presented so that it is more likely to be used by patients, supplementing the insights offered by the information needs taxonomy and providing important guidance for intervention design. Previous studies have measured attributes of consumer health information needs, including level of importance [52], extent of fulfillment [53], amount of information needed [54], and frequency [55], but have largely ignored users' IQ expectations. Our research results suggest that, as an information-related factor that significantly affects system adoption and success [37], IQ should be considered as an important attribute to successfully address patients' information needs.

Theoretically, the results suggest that the model by DeLone and McLean, despite being developed and tested mostly in organizational settings, was effective in guiding the exploration of IQ desired by patients with OC as all quality dimensions specified in the study by Sedera et al [40] were found in our

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data (format was integrated with usability). However, two new dimensions—appropriateness and being sympathetic—emerged from our research. Both dimensions attend to people's emotional states and may be context-specific as most of the participants mentioned that genetic testing occurred during a chaotic and uncertain time when they were busy coping with a cancer diagnosis and dealing with treatment. Efforts are needed to theorize the impact of health information needs and information-seeking contexts on consumer IQ expectations.

Information Delivery

Previous studies have reported that patients with OC are interested in receiving genetic testing–related information through websites, mobile apps, or leaflets [6,12]. We uncovered a wider range of information channel preferences, including interactive technologies (eg, email, patient portals, social media, and smartphone apps), health care providers (through face-to-face conversations, phone calls, and pamphlets), and genetic testing companies and health insurance providers. The inclusion of social media and apps as platforms to receive genetic testing information is a reflection of some patients' interest in hearing other patients' experience with genetic testing, suggesting that peer-to-peer patient education, with its potential to be particularly effective in alleviating fears and strengthening patient empowerment, may be integrated with the dominant provider-led education models to deliver genetic testing information to patients.

The differing preferences expressed by the participants seem to suggest that there may be no one-size-fits-all solution to deliver genetic testing–related information. A hybrid model that uses multiple information channels, media, or platforms and delivers information in both clinical settings and beyond may be needed. For example, leaflets may be distributed in clinics to provide basic and simple genetic testing information to patients, whereas a full-fledged interactive website or app may be created to allow patients to access more advanced and detailed information over the course of their cancer treatment. Web-based communities or social media groups may be created to allow patients to exchange genetic testing–related experiences and information. Simultaneously, health care providers such as nurse navigators and hotline nurses may answer patients' questions by telephone. It is important to note that such solutions should coexist or be integrated with traditional genetic counseling but not replace it.

Similar to most qualitative studies, the results of this study are not quantitatively generalizable in the sense of predicting how many people within a population have certain information needs. However, the rich description of patients' information needs and their IQ and information delivery preferences outlined in this study will help other researchers determine whether the findings are transferable or can be extrapolated to populations with proximal similarities [56]. Toward these ends, the results should be interpreted with the characteristics of the study sample in mind. First, the sample consisted only of women who had undergone genetic testing. The perspectives of women who have not taken genetic tests may provide insights into information gaps experienced by a broader range of patients with OC and shed light on the reasons why genetic testing was not undergone. Second, most of the participants were White and well-educated. Future studies should attempt to include more minority and underrepresented women. Furthermore, the sample did not involve family members, who often serve as delegates to seek information in cancer care [57]. For genetic testing in particular, many patients avail of testing for the sake of their family members [58]. Therefore, understanding family members' information needs may be valuable for intervention design.

Conclusions

Patients with OC have a need for information on various genetic testing–related topics. Genetic counseling alone does not address all of these needs. Interventions that supplement existing genetic counseling are needed. Successful interventions should offer relevant, concise, easy-to-understand, and well-organized (eg, tables and bullet points) information and be available at times and locations needed. Moreover, the information should be appropriate and sympathetic to the cognitive and emotional states of patients with cancer. The patients' preferences for channels or platforms to receive information differed. A hybrid multichannel information delivery model that combines both health care provider–led and peer-to-peer patient education efforts may be most effective in delivering genetic testing–related information to patients with cancer. Future efforts are needed to explore the feasibility of the multichannel information delivery model and its effectiveness in promoting awareness and acceptance of genetic testing among patients and family members and in empowering them in cancer treatment and care.

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Conflicts of Interest

None declared.

Multimedia Appendix 1
Interview and co-design session guide.
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Abbreviations

BRCA: breast cancer gene
CSN: Cancer Survivor Network of the American Cancer Society
IQ: information quality
OC: ovarian cancer
UT: University of Texas

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Original Paper

Web-Based Patient Educational Material on Osteosarcoma: Quantitative Assessment of Readability and Understandability

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Abstract

Background: Patients often turn to web-based resources following the diagnosis of osteosarcoma. To be fully understood by average American adults, the American Medical Association (AMA) and National Institutes of Health (NIH) recommend web-based health information to be written at a 6th grade level or lower. Previous analyses of osteosarcoma resources have not measured whether text is written such that readers can process key information (understandability) or identify available actions to take (actionability). The Patient Education Materials Assessment Tool (PEMAT) is a validated measurement of understandability and actionability.

Objective: The purpose of this study was to evaluate web-based osteosarcoma resources using measures of readability, understandability, and actionability.

Methods: Using the search term "osteosarcoma," two independent Google searches were performed on March 7, 2020 (by AGS), and March 11, 2020 (by TRG). The top 50 results were collected. Websites were included if they were directed at providing patient education on osteosarcoma. Readability was quantified using validated algorithms: Flesh-Kincaid Grade Ease (FKGE), Flesch-Kincaid Grade-Level (FKGL). A higher FKGE score indicates that the material is easier to read. All other readability scores represent the US school grade level. Two independent PEMAT assessments were performed with independent scores assigned for both understandability and actionability. A PEMAT score of 70% or below is considered poorly understandable or poorly actionable. Statistical significance was defined as $P \leq .05$.

Results: Two searches yielded 53 unique websites, of which 37 (70%) met the inclusion criteria. The mean FKGE and FKGL scores were 40.8 (SD 13.6) and 12.0 (SD 2.4), respectively. No website scored within the acceptable NIH or AHA recommended reading level. Only 4 (11%) and 1 (3%) website met the acceptable understandability and actionability threshold. Both understandability and actionability were positively correlated with FKGE ($\rho=0.55$, $P<.001$; $\rho=0.60$, $P<.001$), but were otherwise not significantly associated with other readability scores. There were no associations between readability ($P=.15$), understandability ($P=.20$), or actionability ($P=.31$) scores and Google rank.

Conclusions: Overall, web-based osteosarcoma patient educational materials scored poorly with respect to readability, understandability, and actionability. None of the web-based resources scored at the recommended reading level. Only 4 achieved the appropriate score to be considered understandable by the general public. Authors of patient resources should incorporate PEMAT and readability criteria to improve web-based resources to support patient understanding.

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KEYWORDS

osteosarcoma; patient education; health literacy; web-based health information

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Introduction

Osteosarcoma is a primary malignancy of the bone, affecting 3.4 million individuals globally each year, and is the third most common cancer in the adolescent population [1]. The current treatment for osteosarcoma consists of complete surgical resection coupled with neoadjuvant and adjuvant chemotherapy. Though the introduction of adjuvant chemotherapy in the 1970s has greatly improved survival [2], the diagnosis of osteosarcoma is a significant, life-altering event for patients. In the face of imaging, diagnostic procedures, surgical management, and possible adjuvant treatment, patients may turn to the internet for additional information on their disease and its course.

Once diagnosed with a condition that involves uncertain outcomes, such as osteosarcoma, patients often turn to the internet for additional information. In 2019, approximately 90% of US adults used the internet [3], with an estimated 72% of adults accessing the internet specifically for health information [4,5]. Health literacy is a crucial component of successful health care, with previous studies demonstrating its impact on patient understanding of surgical interventions, adherence to treatment instructions, and even surgical outcomes [6-9]. Alongside growing internet usage in the United States and greater emphasis on shared decision-making, web-based patient educational materials are increasingly recognized as a key component of disseminating health information to improve health literacy in the US population [10]. Recently, the American Medical Association (AMA) and National Institutes of Health (NIH) recommend web-based health information to be written at a 6th grade or lower reading level to be fully understood by the average adult in the United States [11-15].

Most literature assessing patient educational materials has focused on readability measures [6-10,16-21]. However, readability is dependent on the complexity of vocabulary and syntax (linguistics or word order). It provides assessments of written material and is limited in the ability to effectively assess a resource's capacity to convey data such that readers can process and act on the presented information. This limitation has been previously recognized, and the Patient Educational Materials Assessment Tool (PEMAT) was developed to provide more versatile analysis by including two key components of health information: understandability and actionability [19-21]. Understandability is as the ability of readers to process and explain key messages, while actionability is defined as the ability of readers to identify what they can do on the basis of the information presented [21]. While past literature has investigated the readability of web-based osteosarcoma patient educational material, the understandability and actionability has not been previously investigated [18]. The purpose of this study was to use the PEMAT tool to quantify understandability and readability of web-based osteosarcoma patient educational resources, in addition to standard readability algorithms, in order to create a comprehensive analysis that assesses the average patient's ability to read, process, and act on the presented information.

Methods

Education Material Identification

Overview

Education materials were identified using the Google search engine. Google was the search engine of choice because at the time of this study, Google searches comprised 88%-92% of the web-based search market share [16,17]. To best replicate user experience, the authors chose not to use medical or journal portals, as these resources are targeted toward medical professionals and are often not easily accessible to the public. For internal validity, two independent searches were performed on March 7, 2020 (by AGS), and March 11, 2020 (by TRG). The searches were entered to imitate real user experience.

Each reviewer recorded the top 50 websites from their independent search using the term "osteosarcoma." A Google Trends report provides analytics data of the search rate in the United States, including how commonly a specific term is searched. Additionally, various terms can be compared. This Google Trends analysis demonstrated that the term "bone cancer" was searched 4 times more than "osteosarcoma" during the time of this study. However, search results with the term "bone cancer" produced numerous websites unrelated to osteosarcoma, including metastatic lesions, Ewing sarcoma, and chondrosarcoma. Given the specificity of this study, the authors determined it was more appropriate to narrow the search to the desired topic.

Previous analyses of click-through rates report that approximately 70% or more of "clicks" originate from the first 10 search results [22-24], with previous PEMAT studies targeting the top 10 to 50 websites [25-29]. Therefore, each reviewer recorded the top 50 websites using the term "osteosarcoma." After consolidation and removal of the duplicates, websites not meeting inclusion criteria were excluded. Inclusion criteria were websites with the primary content consisting of educational information on osteosarcoma. Exclusion criteria were news articles, primarily audiovisual resources, personal experiences (ie, patient blogs and patient stories on hospital websites), references written for health care professionals (ie, UpToDate, Merck's Manuals), peer-reviewed journal studies, advertisements of a product or service without patient education, articles unrelated to osteosarcoma, and articles not directed at patients as the primary consumer. For example, the initial search included websites related to canine osteosarcoma, which were subsequently eliminated.

Qualitative Characterization

A general tabulation of qualitative website characteristics was performed via qualitative review of the following categories: (1) discussion of operative management, (2) advertisement of a physician or group that provided the described management, (3) discussion of the general background information of the disease (anatomy, pathology, prognosis, and risk factors), (4) discussion of work-up or activities related to diagnosis or preoperative management, (5) discussion of postoperative management, and (6) discussion of complications and risks of operative management. Each website was characterized with a

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"yes" or "no" for each category from (1) to (6), and the characteristics were reported aggregately for across all included osteosarcoma websites. No statistical analysis was performed with the website characteristics for categories (1) to (6).

Statistical Analysis

Readability

The readability of included resources was quantified using objective algorithms: Flesh-Kincaid Grade Ease (FKGE), Flesch-Kincaid Grade-Level (FKGL), Simple Measure of Gobbledygook (SMOG) grade, Coleman-Liau Index (CLI), Gunning-Fog Index (GFI), and Automated Readability Index (ARI). A higher FKGE score indicates that the material is easier to read. All other readability scores represent the US school grade level. These previously validated algorithms were accessed using readability software [25,29-34]. Copyright, references, and weblinks independent of the main text were excluded from the readability analysis.

Understandability and Actionability

Understandability and actionability were assessed using the PEMAT instrument, which is validated by the Agency for Healthcare Research and Quality [19-21]. The PEMAT tool assigns independent understandability and actionability scores for each educational material on a 0%-100% scale. The PEMAT tool includes items 1-19 measurable criteria that span topics such as word choice, organization, use of numbers, content, layout or design, and visual aids. Actionability includes 7 items that assess identifiable action items in the resource, how the reader is addressed, if the reader is provided with explicit steps, tools, calculations, or charts to facilitate completion of an action item. These scores were calculated utilizing the PEMAT criteria with each present criterion receiving 1 point. The number of received points was then divided by the number of possible points and multiplied by 100 [35]. A higher score represents a higher level of understandability or actionability, respectively. The PEMAT developers have established a threshold of 70% as the minimum score required for a web-based resource to be considered actionable and understandable [21].

Understandability and actionability were scored separately for each website by 2 reviewers (MKS and TRG) [19-21,35]. As used previously by the PEMAT developers [19,21], interrater reliability was calculated using Cohen κ.

Search Rank Analysis

Google search ranks were averaged from 2 independently conducted searches. Spearman correlation analysis was performed to assess the correlation between the search rank and its readability, understandability, and actionability. Statistical significance was defined as $P<.05$.

Results

Education Material Identification

A total of 53 unique web-based materials were identified. In total, 37 (70%) websites met the inclusion criteria. In total, 11 websites were excluded as primary literature, 3 were excluded as resources directed at medical professionals, and 2 canine osteosarcoma websites were excluded.

Qualitative Criteria

Of the 37 included resources, all (100%) included background information, and 34 (92%) discussed operative management. The majority of websites (84%) discussed workup and diagnosis, while only 22 (60%) discussed the postoperative course. Risks and complications of operative management were the least included qualitative category, present in only 20 (54%) of the included resources. A total of 10 (27%) websites included an advertisement.

Statistical Analysis

Readability

The mean FKGE score was 40.8 (SD 13.6; Table 1). The mean FKGL, SMOG, CLI, GFI, and ARI scores were 12.0 (SD 2.4), 10.7 (SD 1.9), 14.1 (SD 2.0), 14.2 (SD 2.7), and 11.7 (SD 2.7), respectively. No website scores were at a 6th grade reading level or lower (Figure 1).

Table 1. Flesch-Kincaid Grade Ease score by website. No web-based resource was below the 8th grade reading level.

Score	School level	Interpretation	Websites, n (%)
90-100	5th grade	Easy to read and understand	0 (0)
80-90	6th grade	Easy for conversational English consumers	0 (0)
70-80	7th grade	Fairly easy to read	0 (0)
60-70	8th or 9th grade	Understood by most 13-15–year-olds	4 (11)
50-60	10th or 12th grade	Fairly difficult to read	4 (11)
30-50	College	Difficult to read	20 (54)
0-30	College graduate	Very difficult to read (University graduate level)	9 (24)

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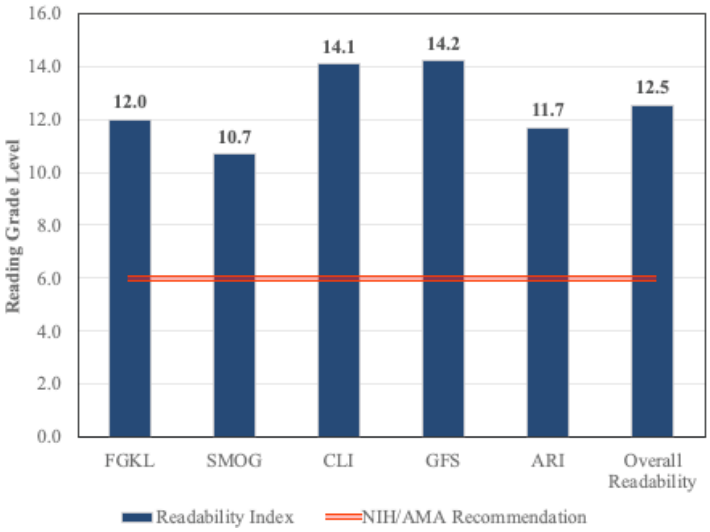
Figure 1. Mean readability index scores. The American Medical Association (AMA) and National Institutes of Health (NIH) recommend health information to be written at a 6th grade or lower reading level (orange line). All mean readability scores exceed this recommended reading level. ARI: Automated Readability Index, CLI: Coleman-Liau Index, FKGE: Flesh-Kincaid Grade Ease, FKGL: Flesch-Kincaid Grade-Level, GFI: Gunning-Fog Index, SMOG: Simple Measure of Gobbledygook.



Understandability and Actionability

Mean understandability and actionability scores were 57.7 (SD 10.7) and 29.1 (SD 22.6), respectively. A total of 4 (11%) and 1 (3%) website met the acceptable understandability and actionability threshold (>70%) for understandability (Figure 2). Interrater reliability demonstrated moderate agreement (Cohen κ=0.78, SD 0.003).

The criteria are listed in Table 2. The most frequently missed understandability criterion was a lack of summary (n=36, 97%), followed by lack of clear titles (n=16, 42%). While 35 (94%) scored well regarding layout and design, only 15 (39%) used visual aids, and only 15 (39%) of those specific sites had visual aids that reinforced rather than distracted from the content. Additionally, only 16 (42%) used common, everyday language and only 21 (58%) appropriately defined medical words. Both understandability and actionability were positively correlated with the FKGE score (ρ=0.55, P<.001; ρ=0.60, P<.001) but otherwise not significantly associated with other readability scores.

Figure 2. Understandability and actionability scores per website. Previous literature reports that a Patient Education Materials Assessment Tool (PEMAT) score of 70% or below is considered poorly understandable or actionable. Four patient educational resources met the understandability threshold, while only one met the actionability threshold. No resources met the threshold for both understandability and actionability.

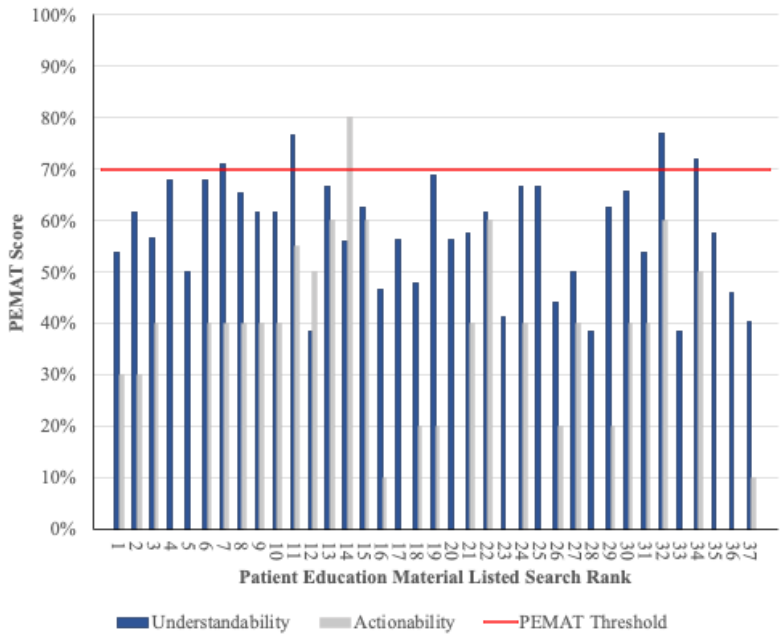


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Table 2. Patient Education Materials Assessment Tool understandability and actionability scoring criteria [21].

Item	Item response	Options
Understandability		
Topic: content		
1	The material makes its purpose completely evident.	Disagree=0, Agree=1
2	The material does not include information or content that distracts from its purpose.	Disagree=0, Agree=1
Topic: word choice and style		
3	The material uses common, everyday language.	Disagree=0, Agree=1
4	Medical terms are used only to familiarize audience with the terms. When used, medical terms are defined.	Disagree=0, Agree=1
5	The material uses the active voice.	Disagree=0, Agree=1
Topic: use of numbers		
6	Numbers appearing in the material are clear and easy to understand.	Disagree=0, Agree=1, No numbers=N/A[a]
7	The material does not expect the user to perform calculations.	Disagree=0, Agree=1
Topic: organization		
8	The material breaks or "chunks" information into short sections.	Disagree=0, Agree=1, Very short material[b]=N/A
9	The material's sections have informative headers.	Disagree=0, Agree=1, Very short material[b]=N/A
10	The material presents information in a logical sequence.	Disagree=0, Agree=1
11	The material provides a summary.	Disagree=0, Agree=1, Very short material[b]=N/A
Topic: layout and design[c]		
12	The material uses visual cues (eg, arrows, boxes, bullets, bold, larger font, and highlighting) to draw attention to key points.	Disagree=0, Agree=1, Video=N/A
Topic: use of visual aids		
15	The material uses visual aids whenever they could make content more easily understood (eg, illustration of a healthy portion size).	Disagree=0, Agree=1
16	The material's visual aids reinforce rather than distract from the content.	Disagree=0, Agree=1, No visual aids=N/A
17	The material's visual aids have clear titles or captions.	Disagree=0, Agree=1, No visual aids=N/A
18	The material uses illustrations and photographs that are clear and uncluttered.	Disagree=0, Agree=1, No visual aids=N/A
19	The material uses simple tables with short and clear row and column headings.	Disagree=0, Agree=1, No tables=N/A
Actionability		
20	The material clearly identifies at least one action the user can take.	Disagree=0, Agree=1
21	The material addresses the user directly when describing actions.	Disagree=0, Agree=1
22	The material breaks down any action into manageable, explicit steps.	Disagree=0, Agree=1
23	The material provides a tangible tool (eg, menu planners and checklists) whenever it could help the user take action.	Disagree=0, Agree=1
24	The material provides simple instructions or examples of how to perform calculations.	Disagree=0, Agree=1, No calculations=N/A
25	The material explains how to use the charts, graphs, tables, or diagrams to take action.	Disagree=0, Agree=1, No charts, graphs, tables, or diagrams=N/A
26	The material uses visual aids whenever they could make it easier to act on the instructions.	Disagree=0, Agree=1

[a]N/A: not applicable.

[b]A very short print material is defined as a material with ≤2 paragraphs and no more than 1 page in length.

[c]These items are only used for audiovisual material.

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Search Rank Analysis

There was no association between readability (P=.15), understandability (P=.20), or actionability (P=.31) scores and Google rank.

Discussion

Principal Findings

This study investigated the understandability, actionability, and readability of web-based resources regarding the diagnosis and treatment of osteosarcoma. Though previous literature has investigated the quality, readability, and social outreach of osteosarcoma materials [18], this is the first study to use the validated PEMAT tool to analyze patient education resources on osteosarcoma. The included osteosarcoma resources scored poorly in all readability measures. Additionally, most of these resources scored under the understandability and actionability standards with only 4 (11%) and 1 (3%) having met the acceptable threshold. Of the 53 resources included, 16 (30%) did not consist of patient education information. These findings confirm existing concerns about the lack of web-based patient material that is readily accessible and consists of high-quality content [21,36-44].

Significance of Web-Based Patient Educational Material

Web-based patient educational material on osteosarcoma is unique, as a large proportion of the patient population consists of adolescents [1,18]. Adolescent internet usage continues to increase, with 45% of teenagers reporting near constant use of the internet, an almost doubling amount since 2015 [5]. Mass media has been cited as a health information resource for teens and is associated with changes in health behavior [45]. However, while adolescents have ready access to the internet, studies have demonstrated unique health literacy challenges within this cohort. Adolescents tend to interact less with the health care system and are therefore less familiar with its navigation [45,46]. Additionally, studies have shown that literacy is a significant challenge for adolescents, with up to 46% reading below the age correlated grade level [45-47]. Therefore, while adolescents are uniquely poised to take advantage of web-based patient resources, providers must be especially mindful of tailoring content to be readable, understandable, and actionable by this younger but technologically savvy patient population.

Readability

Consistent with previous studies, we found osteosarcoma readability scores to be unacceptably above the NIH's and AMA's recommended reading level for public health content [48-50]. This study used common readability index tools and demonstrated that none (0%) of the included websites were written below a 6th grade reading level. In 2016, Cassidy et al [51] reviewed 17 readability studies consisting of orthopedic web-based patient information. They demonstrated that only 0% to 14% had appropriate readability rates using a 6th grade threshold, and only 18% were of the 8th grade reading level [51]. Lam et al [18] found similar results with osteosarcoma educational material and reported that 86% of included websites were written above the 6th-8th grade level [18]. However, rather than using the PEMAT scoring algorithm, they evaluated the qualitative aspects of osteosarcoma websites with the DISCERN instrument [51]. DISCERN criteria score quality on the basis of 16 general questions focused on the patient's opinion of the written material [52]. Overall, DISCERN instrument is used to determine the completeness of the content but does not focus on the reader's ability to understand or act on the material. This study used PEMAT, a validated 24-point scoring system that uses specific variables to evaluate the understandability and actionability of written and visual content.

PEMAT

While readability instruments measure the complexity of the vocabulary and syntax, they do not directly measure the understandability and actionability. Using the reliable and valid PEMAT instrument in this study demonstrated that only 4 (11%) and 1 (3%) included osteosarcoma material met the threshold for understandability and actionability [21]. Additionally, no material met both the understandability and actionability threshold. These scores correspond to those reported in other medical and surgical subspecialities and demonstrate the lack of adequate demonstration of patient education materials on the internet [25,29-31,43,53]. Additionally, there was no association between Google rank and readability, understandability, or actionability; therefore, patients must also be made aware that top ranked websites are not necessarily equivalent with utility or quality.

The osteosarcoma resources were graded using the PEMAT criteria, websites that failed to adhere to the understandability and actionability criteria scored below the PEMAT threshold. In this study, there were several commonly missed understandability criteria across the osteosarcoma websites. The main criteria included missing summaries, lack of visual aids, and unclear titles. Frequently missed actionability criteria included failing to address the patient directly, failing to break down instructions into explicit steps, and failing to provide a tangible action tool such as a checklist. These criteria are valuable aspects of educational material as they optimize the ability for patients to adequately understand content as well as undergo simplistic actions. Missing factors inhibit patient education and can further place the patient at risk for misunderstanding vital material regarding osteosarcoma diagnosis, tests, and treatment modalities [6-9].

To adequately address these deficits, website authors should consider incorporating PEMAT guidelines to ensure the development of patient-appropriate resources [35]. For example, PEMAT guidelines recommend that materials utilize common, everyday language such as "pain killer" rather than "analgesic" [35]. By referencing PEMAT guidelines during the writing process, website authors can create web-based resources that are understandable and actionable.

Limitations

There are limitations of this study that are important to discuss. The top 50 search results are subject to the influence of temporal changes and vary at various times and search locations. The authors cleared all cookies and cache prior to the search to mitigate some variability. The choice of search engine, search

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term, and country of origin can influence the search results. However, the authors utilized the most common search engine with the most specific term: "osteosarcoma." The readability measures can be skewed by certain health care vocabulary. Words including "osteosarcoma" can inherently increase the grade level of the content. Therefore, this aspect may inflate all the grading scores used in this study. However, readability is known to have its limitation in all health care and medical content [54]. Additionally, the subjectivity of the PEMAT grading including implicit bias could not be fully eliminated. To limit this bias and subjectivity, two authors independently performed the grading, which demonstrated agreement with interrater reliability consistent with prior studies utilizing PEMAT [19].

Conclusions

Web-based patient educational material on osteosarcoma scored poorly with respect to readability, understandability, and actionability. None of the web-based resources scored by the AMA and NIH recommended reading level, and only 4 scored above the threshold for what is considered understandable to the general public. Optimization of the most accessible osteosarcoma websites is necessary. Authors of patient resources should incorporate PEMAT and readability criteria to improve web-based resources to support patient understanding.

Conflicts of Interest

None declared.

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Abbreviations

AMA: American Medical Association
ARI: Automated Readability Index
CLI: Coleman-Liau Index
FKGE: Flesh-Kincaid Grade Ease
FKGL: Flesch-Kincaid Grade-Level
GFI: Gunning-Fog Index
NIH: National Institutes of Health
PEMAT: Patient Education Materials Assessment Tool
SMOG: Simple Measure of Gobbledygook

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Original Paper

Efficacy of a Web-Based Psychoeducational Intervention for Young Adults With Fertility-Related Distress Following Cancer (Fex-Can): Randomized Controlled Trial

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Abstract

Background: Threatened fertility following cancer diagnosis in the reproductive age may severely impact emotional and psychosocial well-being in survivorship. Effective web-based interventions for fertility-related distress have been lacking.

Objective: This study aims to test whether the Fertility and Sexuality following Cancer (Fex-Can) intervention is superior to standard care in reducing fertility-related distress and related psychosocial outcomes in young adults with cancer.

Methods: This randomized controlled trial evaluated a 12-week, web-based, automated self-help intervention for fertility-related distress following cancer—Fex-Can Fertility. Individuals were identified via Swedish national quality registries, and those reporting fertility-related distress 1.5 years after diagnosis were invited. A total of 100 women and 24 men (aged 19-40 years) answered self-administered surveys at baseline (T0), directly after the intervention (T1), and 3 months later (T2). The main outcome was fertility-related distress, which was measured by using the 6-dimension Reproductive Concerns After Cancer (RCAC) scale. The secondary outcomes were health-related quality of life (European Organization for Research and Treatment of Cancer Quality of Life Questionnaire), emotional distress (Hospital Anxiety and Depression Scale), fertility-related knowledge, and fertility self-efficacy. In addition, the intervention group (IG) reported self-perceived changes in problems related to fertility after cancer (T1). 2-tailed *t* tests and linear mixed models, including intention-to-treat and subgroup analyses, were performed to compare the effects of the intervention with those of standard care.

Results: Although 62% (31/50) of the participants in the IG stated that their concerns about fertility were fewer after the intervention, there were few statistically significant group differences in the main outcome (RCAC) at T1 and T2. Compared with controls, the IG rated lower distress concerning the dimension child's health at T2 (P=.003; effect size [ES]=0.64). This difference was maintained when adding group and time interactions (intention-to-treat: P=.003; ES=0.58). The IG also had better self-perceived cancer-related fertility knowledge at T1 (P=.05; ES=0.35) and T2 (P=.01; ES=0.42) than the control group. Subgroup analyses based on dose or adherence and baseline RCAC scores did not substantially alter these results. Overall, the use of the web-based program was low.

Conclusions: The Fex-Can intervention had small to moderate positive effects on cancer-related fertility knowledge and distress related to child's health. The lack of group differences in other dimensions of fertility distress and related secondary outcomes

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contrasted with reports on self-perceived improvement after the intervention. The Fex-Can Fertility program may be a useful complement to routine psychosocial support in the clinical care of young women and men with cancer.

Trial Registration: ISRCTN Registry 36621459; https://www.isrctn.com/ISRCTN36621459

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KEYWORDS

cancer; fertility distress; psychoeducation; randomized controlled trial; web-based

Introduction

Background

Physiological and psychological changes following cancer diagnosis and its treatment may have detrimental effects on reproductive health [1,2]. Concerns about fertility and parenthood are among the top unmet care needs of young people diagnosed with cancer, regardless of diagnosis and gender [3]. Reproductive concerns include topics such as uncertainty about one's own fertility potential, concerns about how to tell a current or potential partner about impaired fertility, the fear of recurrence or of one's own health as a barrier to taking care of a family, and distress related to the risk of transmitting cancer genetically to future children [4,5]. Concerns related to fertility and parenthood have been shown to correlate with depressive symptoms [6] and health-related quality of life [3,7-9] in young adults diagnosed with cancer, especially when there is an unfulfilled wish for a child [3,8,10]. Previous research suggests that unmet information needs regarding reproductive health constitute a central aspect negatively affecting the quality of life of women and men diagnosed with cancer in the reproductive age and contribute to fertility-related distress [7]. Intervening with reliable, relevant, and timely information and psychoeducation should, therefore, be the first step toward preventing or alleviating fertility distress. Fertility distress has been studied in qualitative and quantitative research [3,11], and self-administered questionnaires have been developed to measure the phenomenon and study its relationship with other psychosocial variables, such as depressive symptoms and health-related quality of life [4,10,12].

Psychosocial interventions for cancer survivors, which may or may not include web-based components, often have a broad scope [13] and are referred to as *survivorship care plans* [14], self-management interventions [15], or multidimensional programs [16,17]. There is a shortage of interventions targeting both medical and psychosocial concerns regarding fertility and parenthood following cancer. A systematic review of fertility-related psychological distress following cancer reported only 3 psychological interventions [3]. The only web-based intervention study in the field of fertility after cancer that we know of was limited to an educational focus and a single diagnosis (women with breast cancer). The intervention consisted of educational modules aiming to raise participants' knowledge about reproductive health, a web-based bulletin board (discussion forum), and the possibility of interacting with researchers. The study had a noncontrolled, pre-post design and was published as early as 2010 [18].

In the past decade, eHealth has exploded as a research and clinical discipline, and the number of psychosocial and psychological interventions has increased. Several reviews have pointed out the complex nature of eHealth interventions and the challenges involved in their testing and implementation [19-21]. For example, there is limited evidence on dose and adherence measures [22]. The internet is a suitable arena for reaching people in remote areas and approaching private issues; therefore, web-based interventions seem ideal for the topic of fertility and parenthood following cancer. Despite the growing number of web-based interventions in cancer care and survivorship [19,23-26], specific and updated knowledge on the potential of treating fertility-related distress over the internet remains scarce [27].

It has been suggested that to be effective, complex eHealth interventions need to be underpinned by an explicit theoretical framework reflected in the proposed behavior change methods [13,28] and in the choice of outcomes [29]. It has been pointed out that interventions for cancer survivors often lack a theoretical framework and are heterogeneously designed, precluding a thorough evaluation of their working mechanisms [30]. To extend the evidence base, the Fertility and Sexuality following Cancer (Fex-Can) intervention was developed in a participatory process engaging former patients with cancer as research partners [31]. The intervention was conceived in line with the holistic framework for eHealth intervention development [32] and underpinned by the tenets of the self-determination theory (SDT) [33]. According to the SDT, there are 3 universal basic psychological needs—competence (feeling capable), relatedness (feeling connected to others), and autonomy (feeling able to act according to one's inner will). To achieve sustained behavior change and general psychological well-being, all basic needs must be satisfied [29,33]. Therefore, an intervention designed to make participants feel more competent, related to others, and autonomous in relation to decisions surrounding one's fertility was presumed to be effective in the long term. Self-efficacy is presumed to be a proxy measure for competence [34], as it includes not only confidence in knowledge but also the perceived ability to handle actual situations [35]. This confidence in one's capability has been suggested as a mediator for making informed choices and finding motivation for sustainable behavior change in cancer survivorship [36].

The intervention went through feasibility testing [37] and was deemed suitable for the intended target population: women and men aged 19 to 41 years with a recent history of one of the following diagnoses: breast, gynecologic or testicular cancer, lymphoma, or central nervous system tumors.

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Objectives

The aim of this study is to test the efficacy of the Fex-Can intervention in reducing fertility-related distress and psychosocial outcomes in young adults with cancer.

The specific research questions are as follows:

1. Is the Fex-Can Fertility program superior to standard care in reducing fertility distress directly after the end of the program and 3 months later?
2. Does the Fex-Can Fertility program increase fertility self-efficacy and fertility-related knowledge, reduce emotional distress, or improve health-related quality of life compared with standard care?
3. Do baseline levels of fertility distress predict the effect of the program over time?
4. Does dose, that is, the uptake and adherence to the program, influence the change in fertility distress ratings over time?

Methods

Trial Design

The Fex-Can project encompasses a national cohort study [38] with an embedded randomized controlled trial (RCT) including participants with self-reported distress or dysfunction at baseline [39]. The Fex-Can web-based psychoeducational program was offered in two versions—Fex-Can Sex and Fex-Can Fertility, with the latter being evaluated in this study. A detailed description of the study design is available in 2 published study protocols [38,39] and is briefly described in next sections. The Fex-Can Fertility trial is reported here by combining the Template for Intervention Description and Replication checklist [40] with guidelines for eHealth interventions [41] and social and psychological interventions [42], which are both extensions of the original 2010 CONSORT (Consolidated Standards of Reporting Trials) statement for reporting randomized trials [43]. The trial was registered on January 25, 2016 (trial number: 36621459).

Sample

The sample was drawn from a cohort of 1499 individuals diagnosed with breast, cervical, ovarian, or testicular cancer; lymphoma; or central nervous system tumor between 2016 and 2017, approximately 1.5 years before the start of the study. The time frame was chosen to approach people who were likely to have finished primary treatment but were still close enough to diagnosis to be in need of psychosocial support. Eligible participants were identified using Swedish national quality registries, and all people in the intended age bracket (18-39 years at diagnosis) were approached for a longitudinal cohort study, with a letter containing a survey sent to their population registration address. The survey could be completed either on paper or via the web and included written informed consent. Individuals reporting fertility distress at the baseline assessment were invited to the Fex-Can Fertility trial and had to send a signed form back, granting their consent to participate in the RCT.

Eligibility

Respondents scoring ≥4 on at least 1 subscale of the Reproductive Concerns After Cancer (RCAC) scale [4] were eligible for the RCT.

Allocation

Allocation (1:1 ratio) to either the intervention group (IG) or control group (CG) was performed by an external statistician uninvolved in the data collection process by stratified block randomization, taking into account sex and diagnosis. Owing to the design of the intervention, a placebo condition was not possible and neither participants nor researchers could be blinded to the group allocation. Participants were considered lost to follow-up only if they, for any reason, did not return the postintervention questionnaires; therefore, no pattern of attrition was determined after randomization. The flow of participants is summarized in Figure 1.

The sample size was estimated to be 128 individuals needed at follow-up, to obtain statistically significant results, assuming 80% power, medium effect size (ES; 0.5), and a significance level set at .05. As the attrition rate between baseline and first follow-up was expected to be around 15%, we aimed to include 210 participants at baseline.

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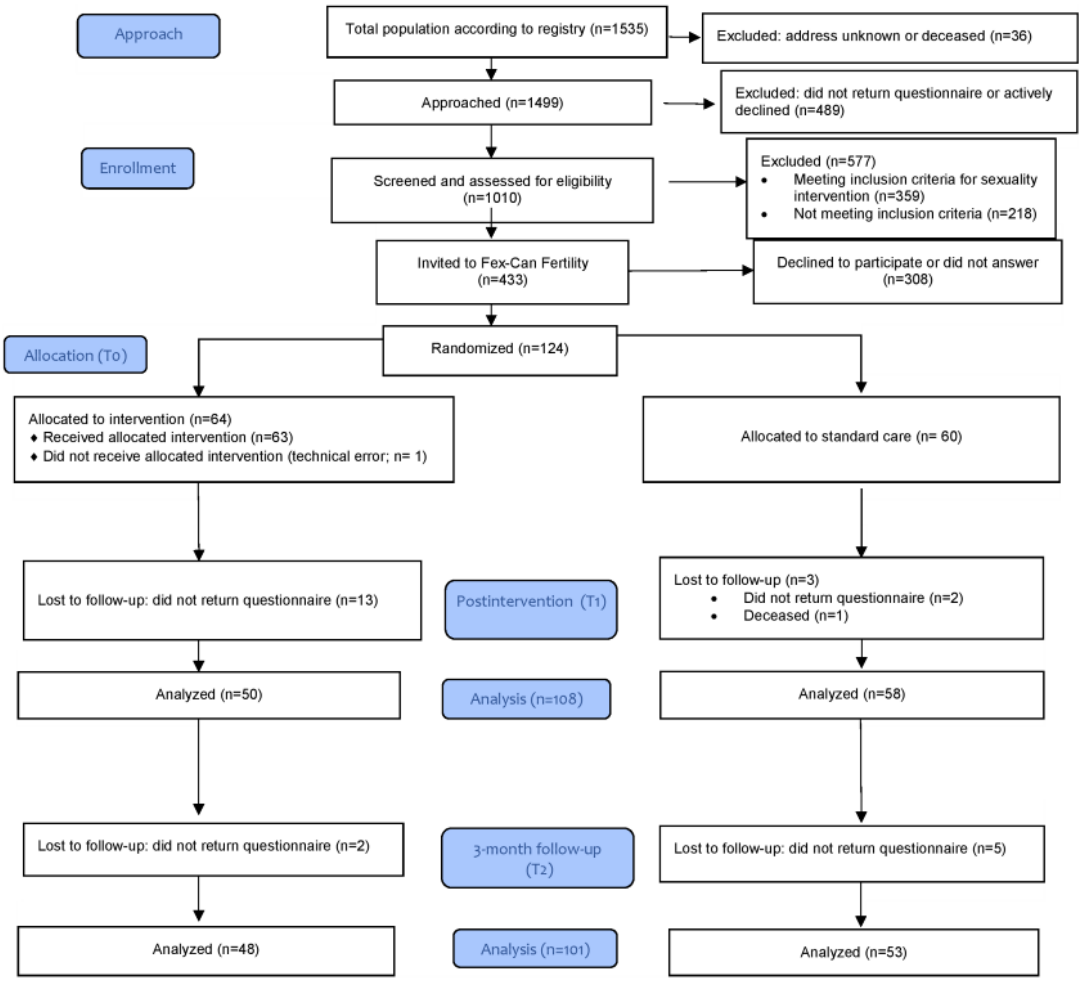
Figure 1. Flow of participants—CONSORT SPI-2018 (Consolidated Standards of Reporting Trials Statement for Social and Psychological Interventions) flow diagram.



Intervention

The intervention was a 12-week, web-based psychoeducational program. The Fex-Can Fertility program was organized in 6 successive modules with informational material, texts, and exercises aiming at developing competence and facilitating behavior change through a sound balance between change and acceptance strategies. The modules covered known aspects of fertility distress [4] and were entitled *Fertility after cancer, Handling anxiety, Trying to have children after cancer, My own health and my child's health, Not being able to have biological children*, and *Relationships*. Contents are described in detail in a doctoral thesis aiming for a process and outcome evaluation of the Fex-Can Fertility intervention [44]. The design, content, and mode of delivery were conceived to facilitate the satisfaction of participants' basic needs according to the SDT [33]. It was

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assumed such theoretical orientation would enhance positive health outcomes such as self-efficacy and health-related quality of life [29]. Nuanced information and reliable facts were intended to leverage participants' competence. Written and filmed survivor stories, as well as interactive quizzes and a discussion forum, were included with the goal of helping participants find strategies to handle their concerns surrounding fertility and family building after cancer by strengthening autonomy and relatedness. The development, design, content, and structure of the intervention have been described in detail in previous studies [31,37,44]. The discussion forum was moderated by one of the research partners [31] and by a member of the research team with clinical expertise in psychology or nursing. Adherence was defined using quantitative activity parameters retrieved from website system data.

Control

The control condition was standard care, which may or may not have included fertility-related support and scheduled contacts with health care, depending on the diagnosis and treatment.

Data Collection

Sociodemographic and Clinical Data

Participants were assessed on outcome measures and sociodemographic variables via a self-administered survey on the following three occasions: baseline (T0), directly after the intervention (T1), and 3 months later (T2). In addition, treatment intensity according to an adapted version of the intensity treatment rating scale [45,46] was assessed using the National Cancer Quality Registry data.

Main Outcome Measure (Fertility Distress)

The RCAC scale was developed for women in the United States with various cancer diagnoses [4] and has been validated for women in China [47] and Sweden [48] and for men in the United States [12]. The scale consists of a total score and six 3-item dimensions related to fertility, pregnancy, and parenthood after cancer: fertility potential (concerns about one's ability to become a biological parent), partner disclosure (concerns related to telling a partner about possibly impaired fertility), child's health (concerns for a biological child's health in relation to the parent's previous cancer diagnosis and treatment, specifically genetic risks), personal health (concerns related to fear of not being able to or living long enough to raise a child), acceptance (the extent of reconciliation with not being fertile or not having biological children), and becoming pregnant (concerns related to efforts involved in achieving a pregnancy). Responses are given on a 5-point scale ranging from strongly disagree (1) to strongly agree (5), where higher scores indicate higher levels of concern. The mean of the total score and the mean scores for each of the 6 dimensions, as recommended in a validation study of the RCAC [49], were used as primary outcomes for the Fex-Can Fertility trial.

Secondary Outcome Measures

Health-Related Quality of Life

Health-related quality of life was measured using the validated [50] summary score (range 0-100) of the European Organization for Research and Treatment of Cancer Quality of Life Questionnaire (version 3.0), which is a generic instrument developed for clinical trials regardless of cancer type [51].

Emotional Distress

The Hospital Anxiety and Depression Scale is a widely used scale measuring anxiety (7 items) and depression (7 items), validated for use in patients with cancer [52]. Scores are given on a numbered Likert scale ranging from 0 to 3, and for each subscale, a total score of 0 to 21 (with higher values indicating more anxiety or depressive symptoms) is calculated.

Fertility Self-efficacy

Perceived confidence in one's ability to manage situations and emotions related to the threat of infertility was measured using a study-specific questionnaire based on previous research [53,54], including 6 items with statements such as "I feel confident that I can tell other people I'm concerned about my reproductive ability." All the items are available in Multimedia Appendix 1. Answers were given on a 4-point Likert scale, with alternatives ranging from completely disagree (1) to completely agree (4). Exploratory factor analysis (data not shown) indicated that one of the items was poorly correlated with the others. The mean score was calculated for the 5 remaining items, with higher values indicating higher levels of fertility-related self-efficacy.

Fertility-Related Knowledge

The perceived level of knowledge concerning fertility issues was measured using a study-specific questionnaire developed from previous research [18], which consisted of 10 items. Answers were given on a 4-point Likert scale, with alternatives ranging from completely disagree (1) to completely agree (4). Exploratory factor analysis (data not shown) of the total cohort of eligible participants indicated that it was suitable to divide the scale into two domains: one for general fertility-related knowledge (4 items) and the other for cancer-related fertility knowledge (6 items). Items included statements such as "I have good knowledge regarding the menstrual cycle and when a pregnancy can occur" (general fertility knowledge) and "I have good knowledge regarding the effect of cancer and cancer treatments on reproductive ability" (cancer-related fertility knowledge). All the items are available in Multimedia Appendix 1. The means were calculated for each subscale, with higher mean scores indicating better perceived knowledge.

Postintervention Evaluation Survey

At T1, participants who had been randomized to the IG were presented study-specific items concerning their experience of the program. Specifically, they were asked to rate their own perceptions of how their problems regarding having children after cancer had changed compared with before participating in the program. Answers were given on a 7-point Likert scale (*Improved a lot*, *Improved*, *Improved a little*, *Did not change*, *Worsened a little*, *Worsened*, or *Worsened a lot*).

Data Analysis

Data were analyzed using descriptive and inferential statistics. Statistical analyses were performed by external statisticians on blinded data. Missing data were treated as follows: for single items that were missing, we imputed according to the individual's mean on the scale, provided half or more of the

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items had been answered. We chose not to impute for individuals where the entire scale was lacking (1-3 participants per group). *t* tests (2-tailed) were used to determine any significant differences between the IG and CG at baseline (T0), directly after the 12-week intervention (T1) and 3 months later (T2). A *P* value inferior or equal to .05 was considered statistically significant. Clinically important changes were calculated using Cohen *d* [55] for ESs, where the difference between the IG and CG mean scores was divided by the pooled baseline SD [56]. ESs of 0.2 to 0.5 were considered small, 0.5 to 0.8 was considered medium, and >0.8 was considered large [55].

Linear mixed models were then used to analyze possible changes over time within and among the treatment groups on the main outcome measure. Mixed models consider the potential dependence of repeated observations within participants and compensate for missing data without the need for imputation [57]. The mixed models included a participant-specific random intercept. The primary end point was T0 (baseline). All available data were used, and the analysis was based on the intention-to-treat principle. In all, 2 types of subgroup analysis were performed. First, for each dimension, participants were assigned to either *high RCAC* (≥4) or *low RCAC* (<4) on the subscale mean at baseline. In the second subgroup analysis, participants were stratified based on three levels of adherence to the program: high, low, and control. High activity was defined as having opened at least half of the modules and spent a total of at least 20 minutes on the website (general activity) plus one of the following: having spent ≥3 minutes in the discussion forum, written a post in the forum, or answered ≥50% of the quizzes (interactivity). All participants who did not meet these criteria were categorized as having *low activity*, which could also include not having logged on to the program at all. For the linear mixed models, ESs were calculated when possible by dividing the point estimate of the group difference by the residual variance. Data were analyzed using SPSS (version 26; IBM Corporation) and Stata (version 16; StataCorp LLC).

Ethical Considerations

This study was approved by the Regional Board of Ethics in Stockholm (permit numbers: 2013/1746-31/4, 2014/224-32, and 2017/916-32) and performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Results

Participants

Eligible participants were persons aged 19 to 40 years, approximately 1.5 years after diagnosis with selected cancer types and reporting elevated levels of fertility distress in a population-based survey. Of the 433 eligible participants approached, 124 (28.6%) agreed to participate. The final sample consisted of 124 individuals, 24 (19.4%) men and 100 (80.6%) women. One participant was assessed at baseline but was excluded from follow-up due to technical failure. Participant characteristics, including sociodemographic and clinical variables, are summarized in Table 1.

Randomization resulted in 64 patients in the IG and 60 in the CG. The attrition was lower than anticipated in the power calculation. At follow-up, of 124 participants, there were 108 (87.1%) and 101 (81.5%) responses from the IG and CG at T1 and T2, respectively (Figure 1).

At baseline, there were no statistically significant differences between the IG and CG in background variables or outcome measures. Breast cancer was the most common diagnosis among participants. Most of the participants had a partner, were working as their main occupation, and had a university or college level of education. Approximately half (29/64, 45% in the IG and 35/60, 58% in the CG) of the participants already had biological children. More than half of the participants (66/124, 53.2%) had received treatments that were very or most intensive or extensive.

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Table 1. Demographic and clinical characteristics recorded at the baseline assessment (T0; N=124).

Characteristics	Intervention group (n=64)	Control group (n=60)
Sex, n (%)		
Men	13 (20)	11 (18)
Women	51 (80)	49 (82)
Age (years), median (range)	33 (20-41)	34 (19-40)
Country of birth, n (%)		
Sweden	54 (84)	51 (85)
Another European country	3 (5)	6 (10)
Outside Europe	7 (11)	3 (5)
Educational level, n (%)		
University	39 (61)	34 (57)
High school	20 (31)	19 (32)
Secondary school or other	5 (8)	7 (12)
Main occupation, n (%)		
Working full-time or part-time	42 (66)	44 (73)
Student	4 (6)	4 (7)
On sick leave	17 (27)	11 (18)
Other (eg, unemployed or full parental leave)	1 (2)	1 (2)
Diagnosis, n (%)		
Breast cancer	26 (41)	26 (43)
Brain tumor	8 (13)	6 (10)
Cervical cancer	10 (16)	12 (20)
Lymphoma	11 (17)	8 (13)
Ovarian cancer	3 (5)	2 (3)
Testicular cancer	6 (9)	6 (10)
Ongoing antitumoral treatment (self-reported), n (%)		
None	40 (63)	39 (65)
Chemotherapy	3 (5)	2 (3)
Radiation	1 (2)	2 (3)
Hormonal treatment	19 (30)	17 (28)
Other (eg, antibodies)	6 (9)	4 (7)
Treatment intensity[a], n (%)		
Level 1: least intensive or extensive treatment	10 (16)	11 (19)
Level 2: moderately intensive or extensive	20 (33)	13 (22)
Level 3: very intensive or extensive	29 (4)	33 (56)
Level 4: most intensive or extensive	2 (3)	2 (3)
Partner, n (%)		
Partnered	50 (78)	52 (88)
Nonpartnered	14 (22)	7 (12)
Parenthood status, n (%)		
Live with children	30 (47)	38 (63)
Had biological children before onset of cancer	29 (45)	35 (58)
Became a parent after cancer	3 (5)	2 (3)

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[a]According to the adapted version of the intensity treatment rating scale.

Use of the Intervention

Of the 64 participants who were randomized to the IG, 21 (33%) reached the level of use defined as *high activity*. Among the remaining 43 participants, 33 (77%) had a lower activity level and 10 (23%) had not logged on to the website at all. With regard to the activity in the discussion forum,17% (11/64) of the participants had made at least 1 posting and 31% (20/64) of the participants had been actively reading the posts for >3 minutes.

Differences Among Groups After Intervention

Primary Outcome

Linear mixed models using a random intercept and based on intention to treat were conducted to study the effects of time and group on the evolution of the main outcome measure. The results are presented in Table 2.

In intention-to-treat analyses, *child's health* was the only dimension in which a significant group difference was detected. The IG had a decrease in scores (ie, reported fewer concerns), and the CG had a slight increase in scores (more concerns) over time (Figure 2; Table 2). At T2, the difference was significant with a moderate ES (*P*=.003; ES=0.576).

Including RCAC baseline scores and activity in the program did not substantially change the results and did not produce any clear pattern (data available in Multimedia Appendices 2-5).

Table 2. Difference in mean values between groups over time (linear mixed models with random intercept: group and time interaction; intention-to-treat: intervention group [IG] vs control group [CG]).

Outcome measure (RCAC[a]; range 1-5) and group	T0 (baseline)	T1 (directly after the intervention)			T2 (3 months later)		
	Value, mean	Value, mean (95% CI)	*P* value	Effect size (Cohen *d*)	Value, mean (95% CI)	*P* value	Effect size (Cohen *d*)
Total mean score			.30	0.20		.22	0.24
IG	3.33	3.17 (3.01-3.32)			3.07 (2.90-3.23)		
CG	3.29	3.28 (3.13-3.43)			3.20 (3.05-3.36)		
Fertility potential			.19	0.25		.26	0.22
IG	3.83	3.50 (3.21-3.78)			3.40 (3.11-3.69)		
CG	3.95	3.76 (3.48-4.04)			3.63 (3.35-3.92)		
Partner disclosure			.42	−0.15		.38	−0.17
IG	3.24	3.15 (2.87-3.44)			2.99 (2.69-3.29)		
CG	2.95	2.99 (2.71-3.27)			2.80 (2.51-3.09)		
Child's health			.11	0.30		.003	0.576
IG	3.25	3.17 (2.86-3.47)			2.91 (2.60-3.22)		
CG	3.49	3.52 (3.22-3.82)			3.59 (3.28-3.90)		
Personal health			.46	0.14		.74	0.06
IG	3.30	3.25 (2.99-3.50)			3.22 (2.96-3.49)		
CG	3.36	3.38 (3.13-3.64)			3.29 (3.03-3.55)		
Acceptance			.64	−0.09		.75	−0.06
IG	3.12	2.98 (2.66-3.31)			2.86 (2.53-3.19)		
CG	2.88	2.87 (2.55-3.20)			2.79 (2.45-3.12)		
Becoming pregnant			.46	0.14		.61	0.10
IG	3.25	3.02 (2.80-3.25)			3.10 (2.86-3.34)		
CG	3.10	3.14 (2.93-3.36)			3.18 (2.96-3.41)		

[a]RCAC: Reproductive Concerns After Cancer.

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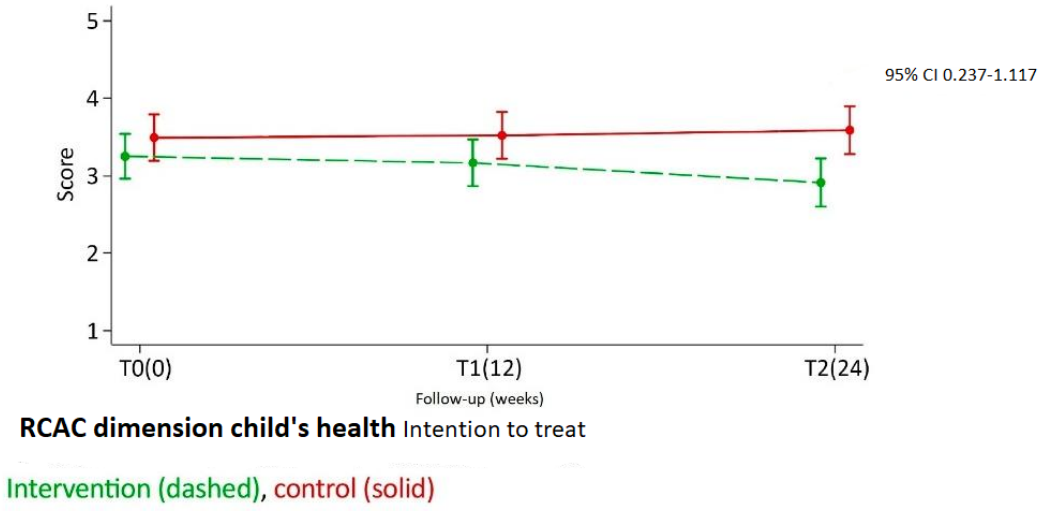
Figure 2. Group differences in dimension 3 (child's health). RCAC: Reproductive Concerns After Cancer.



Secondary Outcomes

There was a significant difference between IG and CG on the secondary outcome cancer-related fertility knowledge, where participants in the IG had better self-rated knowledge than controls at both follow-up points (T1: mean score 2.81 vs 2.54; *P*=.05; ES=0.35 and T2: mean score 2.75 vs 2.38; *P*=.01; ES=0.42). For all other outcome measures, there were no statistically significant differences between groups at T1 or T2 (Table 3).

Table 3. Mean group difference on secondary outcome measures at baseline, after the intervention, and 3 months after the intervention (N=124).

Outcome subscale (range)	T0 (baseline)		T1 (12 weeks; directly after the intervention)				T2 (24 weeks; 3-month follow-up)			
	IG[a] (n=64), mean (SD)	CG[b] (n=60), mean (SD)	IG (n=50), mean (SD)	CG (n=58), mean (SD)	*P* value[c]	Effect size[d]	IG (n=48), mean (SD)	CG (n=53), mean (SD)	*P* value[c]	Effect size[d]
HADS[e] anxiety (0-21)	9.84 (4.45)	8.58 (4.40)	8.67 (4.59)	8.40 (4.46)	.76	0.06	8.71 (3.77)	7.73 (4.58)	.24	0.22
HADS depression (0-21)	5.35 (3.61)	4.76 (4.01)	5.61 (3.85)	4.36 (3.62)	.09	0.33	5.24 (3.87)	3.96 (4.05)	.11	0.33
EORTC-QLQ-C30[f] sum score (0-100)	73.34 (16.80)	76.08 (18.58)	75.06 (17.00)	77.35 (19.40)	.52	0.13	78.01 (16.28)	79.59 (18.15)	.65	0.09
Fertility self-efficacy (1-4)	3.09 (0.75)	3.30 (0.64)	3.21 (0.66)	3.20 (0.66)	.95	0.01	3.13 (0.78)	3.31 (0.67)	.24	0.26
Fertility knowledge										
General (1-4)	3.58 (0.55)	3.50 (0.74)	3.51 (0.68)	3.49 (0.75)	.87	0.03	3.55 (0.57)	3.50 (0.59)	.69	0.07
Cancer related (1-4)	2.65 (0.82)	2.60 (0.73)	2.81 (0.70)	2.54 (0.70)	.05	0.35	2.75 (0.76)	2.38 (0,7)	.01	0.48

[a]IG: intervention group.

[b]CG: control group.

[c]*t* test (2-tailed).

[d]Cohen $d = (\text{mean}_2 - \text{mean}_1)/\text{baseline SD}_{\text{pooled}}$.

[e]HADS: Hospital Anxiety and Depression Scale.

[f]EORTC-QLQ-C30: European Organization for Research and Treatment of Cancer Quality of Life Questionnaire.

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Self-perceived Change in Problems Regarding Fertility After Cancer

At the postintervention evaluation (T1), participants in the IG completed a single item on self-perceived change in problems regarding fertility after cancer. Most of those who completed T1 (31/50, 62%) thought their problems had been alleviated: 30% (15/50 participants) improved a little, 22% (11/50 participants) improved, and 10% (5/50 participants) improved a lot. Of the 50 participants, 18 (36%) felt that their problems had not changed and 1 (2%) experienced a worsening situation and commented that this was not because of the program.

Discussion

Principal Findings

This study invited young adults with cancer who had reported fertility distress in a population-based survey to test the efficacy of a psychoeducational intervention, the Fex-Can Fertility program. This study aimed to determine the capacity of this web-based, self-help program in alleviating fertility distress as measured using the RCAC scale. Assessment at the 3-month follow-up after the end of the program showed significant differences in one out of six dimensions of the RCAC scale, child's health, where the IG had less distress than the CG. Regarding the secondary outcome of cancer-related fertility knowledge, the IG reported better knowledge than the CG at both the directly postintervention and at the 3-month follow-up. ESs were small to moderate, with a more pronounced effect at the 3-month follow-up. Subgroup analyses assessing the possible interaction effect of time and group, adherence, and baseline RCAC scores on the main outcome measure did not substantially alter the results.

Comparison With Previous Work

The results indicating a moderate effect on distress related to genetic risks for offspring and knowledge about fertility after cancer were expected, in the sense that the program contained clear information on these topics. Previous research has found that patients value reliable information and honest communication in health care [58]. One explanation for the effect on the dimension child's health may be that the program provided information that for most cancer diagnoses, the genetic risks involved were small and the amount of uncertainty was low. This knowledge probably calmed participants' worries more than, for instance, learning about the impact of treatment on fertility. The fertility potential is difficult or impossible to exactly predict at the individual level [2]. Similarly, the possibilities of accessing and succeeding with assisted reproductive technologies are concerns where the program might have sustained, or even introduced, a high level of uncertainty. The information provided concerning fertility potential may also have been perceived as negative, as fertility preservation options and assisted reproductive techniques may not have been available for many participants based on female gender, age, diagnosis, or treatment regimen [59]. The same reasoning could apply to, for instance, the dimension personal health, as it contained potentially distressing information on the unpredictable risk of recurrence and potentially new information on harmful, but possibly preventable, late effects of cancer treatment; for example, cardiovascular disease. The intervention showed no effect on fertility self-efficacy, health-related quality of life, and emotional distress.

Adherence and Activity in the Program Were Not Related to Effect

The concept of adherence to eHealth interventions is contested because of a lack of agreement on whether reported measures really refer to use leading to intended effects or simply to use of any kind [22]. In this study, a priori measures of adherence were not established but discussed by the research team at the beginning of the analysis process. To ensure validity, measures of adherence were determined based on the theoretical working mechanisms of the intervention, as suggested in the literature [22]. As in many psychosocial and eHealth interventions, the researchers could not determine an exact cutoff for *high use* a priori, despite previous feasibility testing. There is no theoretical definition of the intended *dose* to achieve a clinically meaningful effect. Whether participants with high use levels also benefited more from the program is, therefore, not completely clear. Qualitative interviews with a subsample of participants in the IG suggested that some individuals who had been relatively inactive found the program, or parts of it, helpful [60]. This may explain why there were no clear results for the models investigating dose or adherence. It could also be that some high-level users became more anxious from the program as they became more aware of treatment-related fertility risks or their own health or because they were in vain searching for comforting information. Some studies suggest that for certain individuals, turning to counseling in health care or looking for support on social media may coincide with an aggravation of distress [61,62]. Considering that the overall use of the program was limited, it can also be questioned whether what was defined a posteriori as *high use* (at least 20 minutes spent on the website, opening half of the modules, and one measure of interactivity in a period of 12 weeks) corresponded to a level of use or intensity that would produce an independent effect.

This study had very small formal dropout rates in the postintervention follow-up, that is, most participants returned questionnaires at both follow-up points. In an intention-to-treat manner, surveys were sent via mail to all patients who were randomized, regardless of their activity level. This means that participants who had not been very active in the program and some who had not even logged on to the website responded to postintervention surveys and were counted as completers alongside their more dedicated counterparts, possibly *washing out* some potential effects of the intervention. However, subgroup analyses based on activity did not show that participants with higher activity benefited more from the program.

Methodological Considerations

Strengths of this study included having a thoroughly prepared, theory-based intervention designed with a participatory approach [31], reaching the whole intended population for eligibility assessment with a validated instrument [38,39], and retaining high response rates throughout the study. However, we wanted to focus on some limitations contributing to why the results must be interpreted with caution.

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RCTs are usually considered the gold standard for scientific evidence. However, in social and psychological interventions, especially eHealth interventions, conditions are not fully controlled, as double-blinding is not possible. The researchers cannot influence what type of accessory support either the IG or the CG has access to, and substantial self-help information is readily available on websites via social or traditional media. This may lead to an inconclusive assessment of intervention effects. Furthermore, there are various sources of bias introduced by design choices, such as not having a set standard for adherence; for example, homework or a minimum assignment for participants. Although evidence for efficacious web-based psychoeducational interventions remains weak [19], reviews on internet-based cognitive behavioral therapy (ICBT) show that therapist-led interventions have larger effects than self-guided programs [63] and are potentially as effective as face-to-face therapy [63,64]. Generally, effective ICBT programs are characterized by a relatively firm structure and limited uptake [65]. The present intervention format was more flexible, and adherence according to the chosen definition did not seem to be associated with an improved effect on the main outcome measure, suggesting that the mechanisms of impact require further investigation and may not be the same as those for ICBT.

To the best of our knowledge, dimensions of the RCAC scale have been used as intervention outcome measures in only one previously published study. A study by Su et al [14] assessed 2 dimensions of the RCAC scale—fertility potential and becoming pregnant—as part of a comprehensive survivorship care plan for women with breast cancer, in which the proportion of participants having improved (moving from >3 to ≤3 on the subscale mean) was statistically significantly larger in the IG than in the CG. In the present study, all dimensions of RCAC were used. It remains unclear whether the instrument is sensitive enough to detect meaningful changes and what the appropriate clinical cutoff level would be. Indeed, when asked in the postintervention survey, 62% (31/50) of the participants in the IG stated that they had improved during the intervention period, but this was not reflected in ratings on the main outcome measure. Active participation in the program was generally low, which may partially explain the lack of group differences. Finally, a contributing factor to not finding more pronounced effects is that despite designing contents of the program to encompass all known aspects of fertility-related distress, the chosen outcome measures may not have adequately captured the change induced by the intervention. Part of the theoretical framework for the intervention relied on efforts to enhance participants' self-efficacy and satisfaction with the basic needs for competence, relatedness, and autonomy. Drawing on the study by Pingree et al [29], we expected that before affecting distal or long-term outcomes, such as quality of life, an intervention may influence intermediate outcome measures, such as basic need satisfaction. The analyses failed to detect statistically significant differences in most outcome measures, including fertility-related self-efficacy. As no measure of basic need satisfaction or other types of motivational measure had been included in the evaluation of the intervention, we were unable to draw conclusions on intermediate outcomes. However, participating in the program did not seem to have produced any adverse outcomes, and most participants stated in both the postintervention survey and qualitative interviews that their distress had been reduced [60], suggesting there was at least some perceived benefit from the intervention.

Conclusions

This web-based psychoeducational intervention for young adults diagnosed with cancer had little overall effect on fertility-related distress. Small to moderate effects could be seen on cancer-related fertility knowledge and the level of concern for future children's health. Further research on the mechanisms of impact is required to determine for whom the Fex-Can program or similar interventions may constitute an appropriate individualized support.

Clinical Implications

The Fex-Can Fertility program could be useful for improving knowledge about fertility and reducing concerns about genetic risks following cancer. The automated, flexible, and partially tailored design of the intervention makes it a convenient tool in clinical care. It appears safe to use because no adverse effects were reported and most participants reported subjective improvement in their concerns.

Acknowledgments

The authors would like to thank all the participants of the Fex-Can (Fertility and Sexuality following Cancer) intervention and the funders.

Data Availability

The software application code and the original data set are available upon reasonable request.

Conflicts of Interest

None declared.

Multimedia Appendix 1
Study-specific instruments.
[DOCX File , 31 KB - cancer_v8i1e33239_app1.docx]

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Multimedia Appendix 2
Significant differences in subgroup analyses based on activity level.
[PDF File (Adobe PDF File), 100 KB - cancer_v8i1e33239_app2.pdf]

Multimedia Appendix 3
Subgroup analyses based on baseline levels of fertility distress.
[DOCX File , 27 KB - cancer_v8i1e33239_app3.docx]

Multimedia Appendix 4
Subgroup analyses based on activity level.
[DOCX File , 54 KB - cancer_v8i1e33239_app4.docx]

Multimedia Appendix 5
Reproductive Concerns After Cancer dimension 5 (acceptance). Subgroup analyses according to baseline levels.
[PNG File , 38 KB - cancer_v8i1e33239_app5.png]

Multimedia Appendix 6
CONSORT eHEALTH Checklist (V 1.6.1).
[PDF File (Adobe PDF File), 117 KB - cancer_v8i1e33239_app6.pdf]

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Abbreviations

CG: control group
CONSORT: Consolidated Standards of Reporting Trials
ES: effect size
Fex-Can: Fertility and Sexuality following Cancer
ICBT: internet-based cognitive behavioral therapy
IG: intervention group
RCAC: Reproductive Concerns After Cancer
RCT: randomized controlled trial
SDT: self-determination theory

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Original Paper

A Smartphone Remote Monitoring App to Follow Up Colorectal Cancer Survivors: Requirement Analysis

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Abstract

Background: Colorectal cancer survivors face multiple challenges after discharge. eHealth may potentially support them by providing tools such as smartphone apps. They have lots of capabilities to exchange information and could be used for remote monitoring of these patients.

Objective: In this study, we addressed the required features for apps designed to follow up colorectal cancer patients based on survivors' and clinical experts' views.

Methods: A mixed methods study was conducted. Features of related apps were extracted through the literature; the features were categorized, and then, they were modified. A questionnaire was designed containing the features listed and prioritized based on the MoSCoW (Must have, Should have, Could have, Won't have) technique and an open question for each category. The link to the questionnaire was shared among clinical experts in Iran. The answers were analyzed using the content validity ratio (CVR), and based on the value of this measure, the minimum feature set of a monitoring app to follow up patients with colorectal cancer was addressed. In addition, a telephone interview with colorectal cancer survivors was conducted to collect their viewpoints regarding a remote monitoring system for colorectal cancer cases.

Results: The questionnaire contained 10 sections evaluating 9 categories of features. The questionnaire was completed by 18 experts. The minimum set of features in the app was identified as patient information registration, sign and symptom monitoring, education, reminders, and patient evaluation ($0.42 < CVR < 0.85$). Features including physical activity, personalized advice, and social network did not achieve the minimum score ($-0.11 < CVR < 0.39$). We interviewed 9 colorectal cancer survivors. Information registration, sign and symptom monitoring, education, and personalized advice were the features with high priority from the survivors' perspectives. Scheduling, shopping, and financial support features were emphasized by survivors in the interview.

Conclusions: The requirement set could be used to design an app for the targeted population or patients affected by other cancers. As the views from both survivors and clinical experts were considered in this study, the remote system may more adequately fulfill the need for follow-up of survivors. This eases the patients' and health care providers' communication and interaction.

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KEYWORDS

eHealth; app; colorectal cancer; survivors; requirements analysis; MoSCoW

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Introduction

Colorectal cancer is one of the most prevalent cancers in the world [1]. Nowadays, with the improvement in health care systems, the number of survivors of this cancer has increased [2]. These survivors face multiple challenges, including a high risk of cancer recurrence. In addition, a high percentage may experience comorbidities from treatment [3].

Financial limitations and pressures caused by the services provided to cancer patients in the health care system result in discharge from the hospital earlier [4]. Therefore, postdischarge care of people with chronic illnesses such as cancer is essential to reduce their readmission [5].

eHealth tools provide a great opportunity to decrease the hospital length of stay and improve care for these survivors. In addition, after discharge, these tools can be used for symptom monitoring, physical activity tracking, psychological issues related to cancer, and nutrition management as well as undergoing a consultation from physicians and health care providers [6].

Positive effects of eHealth interventions on cancer patients' psychological health, appropriate control of their symptoms, communication, knowledge and skills, and quality of life have been reported [4]. The findings have shown cancer survivors' active engagement in their health management [4]. This has led to a major shift from hospital care to informal care at home [7] and patients' attitudes toward self-care and self-management [8].

There is some evidence that recent technologies such as web-based programs [9,10] and smartphone apps [11] can meet information needs related to cancer patients' diagnoses and prognosis management at home. This performance requires that the content and features of these technologies are based on intended users' needs.

Smartphone technologies are rapidly expanding in the health care system due to their availability and ease of use [12] and have a lot of potential for providing access to information, support, and resources from anywhere [13]. However, a limited number of these smartphone apps is devoted to remote monitoring of chronic cancer, especially colorectal cancer, based on the patients' situations to support self-care and making the right decision at each stage of treatment. For example, according to a recent study, 63% of these apps were devoted to diabetes, and only 5% of them related to cancer, and then mainly for information delivery [14].

Consequently, smartphone apps can play an effective role in helping with the follow-up of patients with colorectal cancer, remote monitoring of physical and mental signs and symptoms, and improving health care through patient understanding of what they need to do in each phase via an easy electronic consult with their clinical experts. Therefore, the purpose of this study was to identify and analyze the required features of remote monitoring smartphone apps designed to follow up colorectal cancer survivors with the focus of supporting them after surgery.

Methods

A cross-sectional, mixed methods study was designed to determine the requirements for a smartphone app to monitor colorectal cancer survivors after discharge and was conducted in 2019. The requirements for this smartphone app were gathered from a previous study that investigated eHealth tools for supporting colorectal cancer survivors, by reviewing articles [15-29]. The features of the apps introduced in these articles were extracted and reviewed by a group of medical informatics experts (n=3) and validated by a clinical specialist (n=1, MS). Then, a questionnaire was created containing the requirements based on the extracted features. The questionnaire was generated on an online questionnaire builder platform, and the link to the questionnaire was shared via Telegram messenger in a group including oncological surgeons and related clinical experts.

The questionnaire was composed of 10 parts. Except for the first part that was designed to gather the responders' information, the next 9 parts were designed to obtain the experts` opinions about apps requirements (or the so-called features). The questionnaire structure is shown in Table 1.

For sections 2 to 8 of the questionnaire, items were scaled as 4-choice questions based on the MoSCoW (Must have, Should have, Could have, Won't have) method [30], and at the end of each section, an open question regarding comments on that section was asked.

MoSCoW is a technique used for requirement prioritization. This technique categorizes each requirement into "Must have," "Should have," "Could have," and "Won't have" requirements. The "Must-have" requirements indicate that the feature must be implemented in this version. The "Should have" requirements indicate that the features must be implemented in this version if at all possible. The "Could have" requirements indicate that the features could be implemented if they do not affect any other requirement. The "Won't have" requirements indicate features that are not needed in this version but could be included in the future.

In the next phase, the content validity ratio (CVR) was determined using the formula:

where N is the total number of experts (n=18) and N_e refers to the count of experts that chose "Must have" or "Should have" to consider the features as essential requirements. The threshold was considered based on the nearest original thresholds introduced by Lawshe [31]. In his work, he provided a table containing the minimum required CVR for an item to be selected based on the number of content evaluation panel members. In this study, the threshold was set at 0.42, based on the table by Lawshe [31].

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Table 1. Questionnaire items designed to obtain the experts' opinions about the required features of apps designed to follow up colorectal cancer survivors.

Section codes, section names, and item codes	Items
Section 0: Questionnaire responder information	
Item 0.1	Name (optional)
Item 0.2	Gender
Item 0.3	Expertise
Item 0.4	Work experience duration
Item 0.5	Activity type
Item 0.6	City
Item 0.7	Cell number (optional)
Item 0.8	email address (optional)
Section 1: Patient information registration	
Item 1.1	Sociodemographic information
Item 1.2	Diagnosis and previous surgery information
Item 1.3	Surgery and after surgery information
Item 1.4	Comments on this section
Section 2: Sign and symptom monitoring	
Item 2.1	Weight tracking
Item 2.2	Vital sign tracking
Item 2.3	Symptom tracking
Item 2.4	Side effect tracking
Item 2.5	BMI tracking
Item 2.6	Comments on this section
Section 3: Education	
Item 3.1	Information about cancer
Item 3.2	Common issues for patients
Item 3.3	Information about physical activity
Item 3.4	Information about drugs
Item 3.5	Information about chemotherapy
Item 3.6	Information about nutrition
Item 3.7	Information about rehabilitation
Item 3.8	Information about the treatment process
Item 3.9	Information about postdischarge
Item 3.10	Information about pain management
Item 3.11	Information about emergency issue management
Item 3.12	Other patients' experiences
Item 3.13	Comments on this section
Section 4: Physical activity	
Item 4.1	Goal setting for physical activity
Item 4.2	Physical activity tracking
Item 4.3	Viewing other patients' physical activity progress
Item 4.4	Comments on this section
Section 5: Reminders	

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Section codes, section names, and item codes	Items
Item 5.1	Hospital visit reminders
Item 5.2	Medication reminders
Item 5.3	Comments on this section
Section 6: Personalized advice	
Item 6.1	Online consultation system
Item 6.2	Tailored patient information
Item 6.3	Comments on this section
Section 7: Patient evaluation	
Item 7.1	Quality of life evaluation
Item 7.2	Nutrition evaluation
Item 7.3	Clinician-patient relationship evaluation
Item 7.4	Comments on this section
Section 8: Social network	
Item 8.1	Patients' discussion groups
Item 8.2	Comments on this section
Section 9: Comments	
Item 9.1	Other comments

In the second phase, a one-on-one, phone, semistructured interview was conducted to ask colorectal cancer survivors' opinions about a mobile app's features. The open question and the guide questions were designed and finalized based on the research team's (SMA, SRNK) ideas. The list of colorectal cancer survivors who had already visited the Cancer Institute of Imam Khomeini hospital in Tehran and were discharged during the last year (2019-2020) was prepared, and sampling was conducted randomly. The telephone numbers of all the patients were available, and the researcher called them to ask the designed questions. Unfortunately, 5 calls resulted in the very bad news of the patient's death after discharge. The interview was conducted with the remaining 9 survivors or one of their family members.

The guide for asking the open question was "What features do you think a mobile app needs to help you for better care? Or do you have any health problems that an app could help you in that condition?"

After asking the open question, the survivors were asked to prioritize the feature categories, by scoring each feature category from 1 (the lowest priority) to 4 (highest priority).

The interviews were transcribed, coded, and categorized into themes for qualitative analysis. In the quantitative analysis, to rank the features based on scores, the CVR of each category was calculated, and the threshold was set at 0.78 based on the thresholds by Lawshe (n=9).

Results

Clinical Experts' Information

The questionnaire was completed by 18 experts (3 women, 15 men): 7 were oncological surgeons, 5 were general surgeons, and 3 were clinical oncologists. The others included an internist, a laparoscopic specialist, and one who did not specify his expertise. Most (11/18, 61%) possessed a work experience of 5 years to 10 years. They were working mostly in Tehran, the capital of Iran. Others were working in the other 7 cities in the country. One worked in the 2 cities of Tehran and Khorasan concurrently. The responders' characteristics are shown in Table 2.

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Table 2. Responders' characteristics (n=18).

Characteristics	Value, n (%)
Gender	
Female	3 (17)
Male	15 (83)
Specialty	
General surgeon	5 (28)
Oncology surgeon fellowship	7 (39)
Clinical oncologist	3 (17)
Other	3 (17)
Work experience	
Less than 5 years	3 (17)
Between 5 and 10 years	11 (61)
Between 11 to 15 years	1 (6)
Between 16 to 20 years	1 (6)
More than 20 years	2 (11)
Faculty	
Faculty member	11 (61)
Not faculty member	7 (39)
City	
Tehran	7 (39)
Tehran and Khorasan	1 (6)
Yazd	2 (11)
Mashhad	1 (6)
Gorgan	1 (6)
Gerash	1 (6)
Shiraz	1 (6)
Sanandaj	1 (6)
Dezful	1 (6)
Ahvaz	1 (6)
Isfahan	1 (6)

Requirement Prioritization

The responses to the 4-choice questions based on the MoSCoW method and the CVR for each item are represented in Table 3. All experts had consensus on the "Diagnosis and previous surgery information" and "Surgery and after surgery information" items.

Based on CVR values for each item shown in Table 3 and the threshold (0.42), 21 items should be considered as essential requirements, categorized in 8 main groups.

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Table 3. Responses to the 4-choice questions and content validity ratio (CVR) for each item (n=18).

Item	Must have, n	Should have, n	Could have, n	Won't have, n	CVR
Patient information registration (overall CVR=0.85)					
Sociodemographic information	10	4	4	0	0.56
Diagnosis and previous surgery information	18	0	0	0	1.00
Surgery and after surgery information	18	0	0	0	1.00
Sign and symptoms monitoring (overall CVR=0.42)					
Weight tracking	9	6	3	0	0.67
Vital sign tracking	5	4	7	2	0.00
Symptom tracking	6	4	7	1	0.11
Side effect tracking	16	2	0	0	1.00
BMI tracking	5	7	5	1	0.33
Education (overall CVR=0.58)					
Information about cancer	10	3	4	1	0.44
Common issues for patients	13	2	3	0	0.67
Information about physical activity	6	6	5	1	0.33
Information about drugs	8	5	3	2	0.44
Information about chemotherapy	9	7	2	0	0.78
Information about nutrition	14	3	1	0	0.89
Information about rehabilitation	12	5	1	0	0.89
Information about treatment process	12	1	4	1	0.44
Information about post discharge	13	4	1	0	0.89
Information about pain management	9	5	3	1	0.56
Information about emergency issue management	14	3	1	0	0.89
Other patients experience	5	2	8	3	–0.22
Physical activity (overall CVR=–0.22)					
Goal setting for physical activity	3	4	9	2	–0.22
Physical activity tracking	4	4	8	2	–0.11
Viewing other patients' physical activity progress	4	2	9	3	–0.33
Reminders (overall CVR=0.84)					
Hospital visit reminder	14	3	1	0	0.89
Medication reminder	12	4	2	0	0.78
Personalized advice (overall CVR=0.39)					
Online consultation system	3	9	5	1	0.33
Tailored patient information	7	6	5	0	0.44
Patient evaluation (overall CVR=0.70)					
Quality of life evaluation	10	5	3	0	0.67
Nutrition evaluation	12	6	0	0	1.00
Clinician-patient relationship evaluation	9	4	5	0	0.44
Social network (overall CVR=–0.11)					
Patients' discussion groups	1	7	8	2	–0.11

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Open Question Responses

One specialist commented on section one and stated that "the patient's history with rich data could be helpful for retrospective studies and will increase the importance of the app."

Two other specialists commented on the items of section three. One asked the following question: "To which extent the data will be provided to the patient?" In addition, the other had concerns about sharing other patient experiences by commenting: "the patients' experiences are usually not scientific and have no profound evidence." This expert also commented on the items in section eight, by stating "I disagree with all kinds of opinion exchange with patients. Because unfortunately, most of the time it led to wrong information transfer." Another expert also commented on this section by stating "regular group sessions with clinicians and patients [will be helpful]."

In the last section, 3 experts left their comments. One noted that: "I believe that [an app with these features] are too useful." The other hoped success for the research team. The third addressed 3 issues:

> *(1) regarding patients with colostomy, the app should provide exact information about the bag and notes for changing that; (2) possible emergency after surgery such as fever and infection, gastrointestinal hemorrhage, thromboembolic, and signs and symptoms of recurrence should be informed to the patient; (3) patients have interest in [care] details such as nutrition, physical activity, etc.; in this app, these issues should be noticed.*

Telephone Interviews With Colorectal Cancer Survivors

The survivors' characteristics are shown in Table 4.

The themes extracted from the interviews were accessibility and usability; 2-sided information flow, to inform and get informed; scheduling; shopping; decision-making; social support; and financial support.

Table 4. Colorectal cancer survivors' characteristics (n=9).

Characteristics	Value, n (%)
Gender	
Female	5 (56)
Male	4 (44)
Age (years)	
50-59	3 (33)
60-69	4 (44)
70-79	2 (22)
City	
Tehran	3 (33)
Arak	1 (11)
Sanandaj	1 (11)
Karaj	1 (11)
Elam	1 (11)
Borujerd	1 (11)
Khoramabad	1 (11)
Cancer	
Colon	8 (89)
Rectum	1 (11)
Chemotherapy	
Yes	8 (89)
No	1 (11)

Accessibility and Usability

One survivor mentioned she does not have a smartphone. Another mentioned an older adult survivor could not work with the app, although a member of the family may help and work with the system instead.

2-Sided Information Flow, to Inform and Get Informed

The survivors wanted to be monitored. The app should ask questions about the survivor's health and inform the clinician about the survivor's status. The survivors could ask questions regarding what to do with nutrition, losing weight, high blood pressure, and other challenges. The survivors wanted to know

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what to do in each condition. They need a consultation. They become anxious about unknown side effects. The app should provide information about colostomy bags. In addition, the possibility of sending high-quality lab results to the clinician via an electronic tool was requested.

Scheduling

The survivors complained of the scheduling process, and they requested that the app have features to ease the scheduling process, especially for those who are traveling from cities located far across the country from Tehran where more highly specialized physicians are available.

Shopping

Some survivors complained of the hardness to get and find medications for their cancer and asked for a feature in the app to sell medications and colostomy bags.

Decision-making

One survivor pointed out that the app should log medication intake and the outcomes of taking each medication. This would enable clinicians to make better decisions.

Social Support

One survivor mentioned that the discussion group can provide social support.

Financial Support

Some patients were deeply unhappy about the cost of a colostomy bag, traveling, medication, and even access to smartphone expenses. The app might help them to be connected with donations and charity organizations.

The survivors acknowledged the app helps the survivors, especially those who are in cities other than their clinician's city (in this case, Tehran) because of traveling. Furthermore, they pointed out that such an app is more useful in the COVID-19 pandemic.

The priority of feature categories is shown in Table 5.

Table 5. Responses to the 4-choice questions (n=9).

Item	Highest priority (4), n	Above medium priority (3), n	Medium priority (2), n	Lowest priority (1), n	CVR[a]
Patient information registration	7	1	1	0	0.78
Sign and symptom monitoring	7	2	0	0	1.00
Education	9	0	0	0	1.00
Physical activity	4	3	1	1	0.56
Reminders	5	2	2	0	0.56
Personalized advice	6	2	1	0	0.78
Patient evaluation	3	4	1	1	0.56
Social network	5	1	1	2	0.33

[a]CVR: content validity ratio.

Discussion

Principal Findings

In this study, the requirements for a remote monitoring smartphone app to follow up colorectal cancer survivors were extracted and prioritized using the MoSCoW method and CVR. The proposed questionnaire was completed by 18 specialists, and an interview was conducted with 9 colorectal cancer survivors.

Although a successful app should consider all stakeholders in the design process [32], the patients, as one of the primary stakeholders, should be involved in the design. Thus, it is necessary to listen to and get feedback from patients during the elaboration of the design and prototyping the app. In this study, we addressed the specialists' view of such an app. The findings align with those of previous studies that addressed colorectal cancer survivors' needs, which we discuss in the following paragraphs.

Information about the diagnosis and treatment summary as well as medical and nonmedical needs were shown to be useful for colorectal cancer survivors. Other needs, such as information about late effects and likely issues including fatigue and bowel-related symptoms, could be helpful for them. They also need information about nutrition and general health. A list of recommended tests is also important for survivors. In addition, information about cancer, prognosis, and recurrence of cancer are suitable. Some survivors may prefer to be informed about finding a local health care center for ongoing care, personalized information, and trusted sources of information [33]. Generally, the met and unmet needs of colorectal cancer survivors could be categorized as physical symptoms, emotional, information, and coping strategies [34]. The designed app should address these information needs of the colorectal cancer survivors. However, previous apps designed for cancer survivors have not satisfied all the information needs of these survivors such as psychological support, managing finances, and long-term effects [35]. A comprehensive app addressing these needs, as included in the requirements analysis, might be more supportive for survivors.

The 2 items entitled "Diagnosis and previous surgery information" and "Surgery and after surgery information" were

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added to the questionnaire based on the expert author's (MS) comment. These 2 items had a CVR of 1, which means all experts had consensus on these items. In addition, from the survivors' view, this feature was necessary. "Sign and symptom monitoring" was extremely important from the survivors' point of view, and, similarly, "Side effect tracking" also had a CVR of 1, which might be due to the high rate of colorectal cancer comorbidities and treatment effects, as mentioned before in [3] in which 18% of patients with colorectal cancer experienced at least one comorbidity after their discharge.

The "reminders" domain, including items for hospital visit reminders and medication reminders, had a high score, indicating the experts believed that reminders are important and the app could manage them effectively. In contrast, reminders were not considered essential from the survivors' point of view, although most of them thought reminders were of high priority. One underlying reason may be reflected by one survivor's view that "families are engaged in the health care, and there is no need for reminders."

The education domain, except physical activity training, and the reminder domain could be effectively implemented from the experts' point of view. This domain is considered essential from the survivors' point of view.

Physical activity has been shown to be effective in reducing colorectal cancer mortality [36] and its negative effects [37]. However, the "physical activity" domain in addition to the "Information about physical activity" items in this study did not reach the minimum score to be in the minimum feature set of such a smartphone app. The reason may be related to studies such as [15,24] that showed eHealth is not effective in improving physical activity behaviors of cancer patients. Some survivors mentioned that they could not perform physical activity due to their condition. Perhaps, for younger survivors, this feature would be marked as essential.

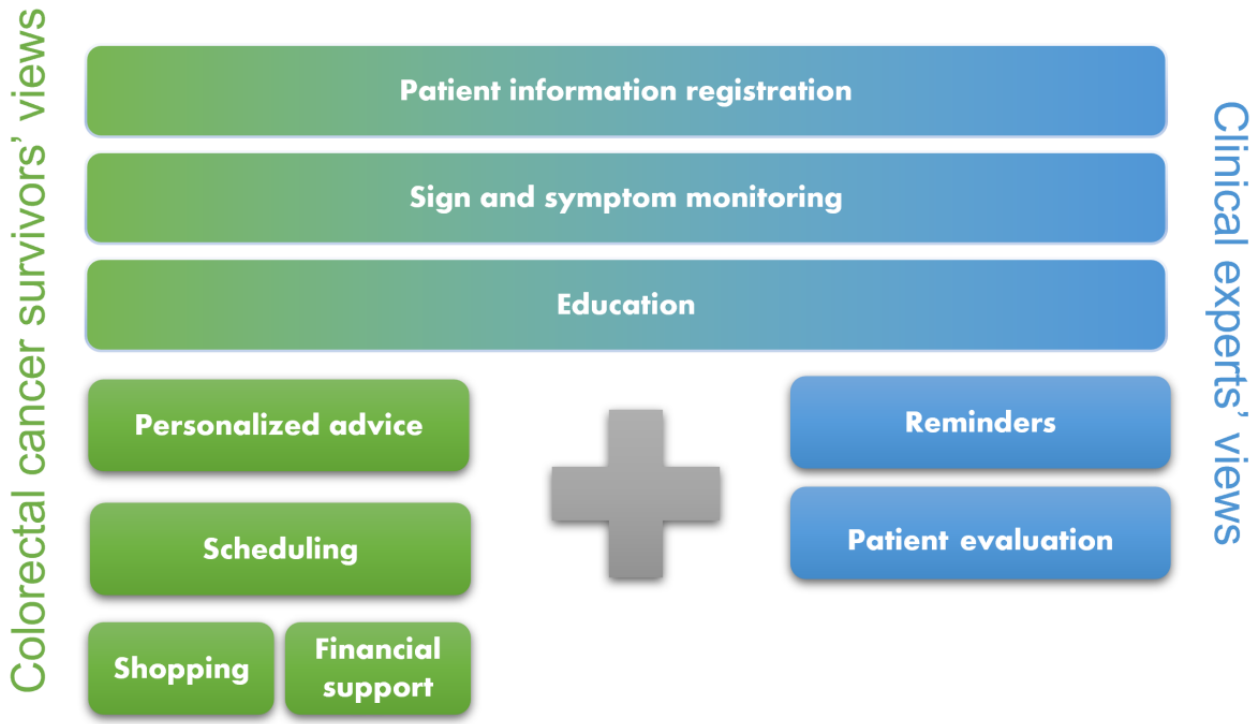
The "patients' discussion groups" domain did not pass the test; this might be represented by the statement of one expert: "the patient's experiences are usually not scientific and have no profound evidence." Thus, this causes the problem of misinformation exchange between patients. Some patients had the same idea about this feature. However, other patients mentioned that it could be useful to know the trajectory of other patients and that it could provide social support.

The "personalized advice" domain was a priority for survivors, although the clinicians did not consider consultations as a high-priority feature. The reason may be the lack of clinicians' time and that the costs are not covered by any party.

As mentioned by one of the experts, training and notes about the use of a colostomy bag should be considered in the app, particularly in the education section. Regarding the other comment from an expert who stated the app could be helpful, it is worth mentioning that his or her comment could be true in general. However, the effectiveness of these apps in the domains of survivors' nutritional status and social support has not been shown [38].

All the high-priority requirements are shown in Figure 1. The requirements were gathered form the combination of the colorectal cancer survivors' and clinical experts' priorities based on the CVR and items mentioned by more than one survivor in the interviews.

Figure 1. Requirements for a smartphone app to remotely monitor and follow up colorectal cancer survivors.



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Overall, the findings suggest that a smartphone app to remotely monitor patients with colorectal cancer after discharge should be designed to include the features of patient information registration, sign and symptom monitoring, education, reminders, patient evaluation, personalized advice, scheduling, shopping, and financial support.

Recently, especially during the COVID-19 outbreak, eHealth interventions have gained more attention from governments and populations. They are showing more encouragement for remote interventions. This could lead to progress in investment in such apps. The suitable design and standardization of features for these apps may help to better provide support to these survivors. In this study, the features of the app were defined based on experts' views; future works could focus on obtaining patients' views and elaborating these features.

Strengths and Limitations

One of the strengths of this study was the provision of a feature set for a remote monitoring and educational app for colorectal cancer survivors after surgery for the first time in the country. In addition, the combination of qualitative questions and quantitative methods and obtaining clinical experts' and survivors' viewpoints is an approach that could also be considered a strength. Another strength is the collaboration between oncological experts and the research team.

The research was conducted in Iran, and the priorities for the features could be different in other countries. Thus, this could be considered a limitation of this study. The sample size could also be considered a limitation.

Conclusion

In this study, the requirements of a remote monitoring smartphone app to follow up colorectal patients were determined by literature review, specialists' confirmation, and survivors' viewpoints. These requirements might help design such an app. Further research should address the generalizability of the feature set in other cancers and the possibility of defining standards for such apps.

Conflicts of Interest

None declared.

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Abbreviations

CVR: content validity ratio
MoSCoW: Must have, Should have, Could have, Won't have

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Original Paper

Health Promotion Among Mexican-Origin Survivors of Breast Cancer and Caregivers Living in the United States–Mexico Border Region: Qualitative Analysis From the Vida Plena Study

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Abstract

Background: Hispanic survivors of cancer experience increased cancer burden. Lifestyle behaviors, including diet and physical activity, may reduce the cancer burden. There is limited knowledge about the posttreatment lifestyle experiences of Hispanic survivors of cancer living on the United States–Mexico border.

Objective: This study aims to support the development of a stakeholder-informed, culturally relevant, evidence-based lifestyle intervention for Mexican-origin Hispanic survivors of cancer living in a border community to improve their dietary quality and physical activity.

Methods: Semistructured interviews with 12 Mexican-origin Hispanic survivors of breast cancer and 7 caregivers were conducted through internet-based teleconferencing. The interviews explored the impact of cancer on lifestyle and treatment-related symptoms, perception of lifestyle as an influence on health after cancer, and intervention content and delivery preferences. Interviews were analyzed using a deductive thematic approach grounded in the Quality of Cancer Survivorship Care Framework.

Results: Key survivor themes included perception of Mexican diet as unhealthy, need for reliable diet-related information, perceived benefits of physical activity after cancer treatment, family support for healthy lifestyles (physical and emotional), presence of cancer-related symptoms interfering with lifestyle, and financial barriers to living a healthy lifestyle. Among caregivers, key themes included effects of the cancer caregiving experience on caregivers' lifestyle and cancer-preventive behaviors and gratification in providing support to the survivors.

Conclusions: The interviews revealed key considerations to the adaptation, development, and implementation of a theory-informed, evidence-based, culturally relevant lifestyle program to support lifestyle behavior change among Mexican-origin Hispanic survivors of cancer living in border communities. Our qualitative findings highlight specific strategies that can be implemented in health promotion programming aimed at encouraging cancer protective behaviors to reduce the burden of cancer and comorbidities in Mexican-origin survivors of cancer living in border communities.

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KEYWORDS

Mexican-origin Hispanics; breast cancer; survivorship; caregivers; border health; lifestyle; diet; physical activity; health promotion; mobile phone

Introduction

Background

Cancer remains the leading cause of death among Hispanic people in the United States [1]. Cancer incidence rates are higher among Hispanic people compared with non-Hispanic White people for obesity-related cancers [1]. Obesity-related cancers have a metabolic etiology that can result in significant comorbidities. Many survivors of cancer experience comorbid conditions that may influence prognosis. Common comorbid conditions include cardiovascular disease, type 2 diabetes mellitus, hypertension, and obesity [2-4] and metabolic diseases that disproportionately affect Hispanic patients [5]. The burden of these comorbidities is higher among Hispanic people [6-8]. Among border-dwelling Hispanic people in the United States, mortality incidence rates exceed those in non-Hispanic White people for all obesity-related cancer types [9], which supports the need for health promotion programs to reduce health disparities in this vulnerable group.

Disparities in health are magnified in populations living along the United States–Mexico border. The border-dwelling population is largely of Mexican origin and is expected to double in size in the coming years [10]. Border communities are medically underserved and experience higher rates of poverty, poorer health outcomes, and lower access to professional health care than the general US population [10,11]. Breast cancer, an obesity-related cancer, is the most commonly diagnosed cancer type and the leading cause of cancer death among Hispanic women [1]. Although it is known that certain health behaviors are cancer protective, such as plant-dominant eating patterns and physical activity [12,13], previous research has shown that cultural health beliefs, social norms, access to resources, and cultural food preferences influence the health behaviors of Hispanic women [14,15]. Of note, compared with non-Hispanic White survivors of breast cancer, Hispanic survivors of breast cancer are more likely to report their health as fair or poor and are less likely to meet diet and physical activity recommendations [16,17].

Currently, there is limited knowledge of the cancer survivorship experience of Mexican-origin women, especially those living along the United States–Mexico border. The Quality of Cancer Survivorship Care Framework developed by Nekhlyudov et al [18] suggests that cancer survivorship quality is influenced by a variety of interrelated care needs, including management of physical and psychosocial symptoms related to treatment, control of comorbid conditions, and adoption of prevention-focused lifestyle behaviors. Health promotion interventions are poorly accessed by Hispanic populations [19,20], potentially because the components of these interventions lack relevance [21,22]. Culturally adapted, stakeholder-informed interventions are more successful in reaching high-risk, underserved populations [23,24]. Barriers to intervention adoption include language accessibility, lack of nutrition education constructed around culturally-based foods, and poor access to resources [22]. As compared with non-Hispanic White women, Mexican-origin Hispanic survivors of breast cancer have been shown to have poorer health-related quality of life and poorer adherence to diet and physical activity recommendations [16,17]; it is important to determine aspects to include in lifestyle interventions that can more appropriately meet the unique needs of Mexican-origin Hispanic survivors of cancer. Stakeholder input is central to this process.

Objectives

Developing and adapting a lifestyle intervention that includes components that are culturally relevant can increase the acceptability of health promotion programs for women living along the United States–Mexico border and more effectively help promote cancer protective behaviors, specifically dietary quality and physical activity, ultimately improving health outcomes. The aims of this qualitative study are to (1) determine the important aspects for developing and adapting a culturally relevant lifestyle intervention for survivors of cancer in border communities and 2) contribute to a greater understanding of lifestyle behavior and survivorship in Mexican-origin breast survivors of cancer and their caregivers.

Methods

Overview

This qualitative study is part of Vida Plena, a community-based participatory research study [25,26] between the Mariposa Community Health Center (MCHC) and the University of Arizona Cancer Prevention and Control Research Network. MCHC is a designated federally qualified health center in Nogales, Arizona, United States, which provides primary care services to nearly half of all residents of Santa Cruz County, Arizona, United States. Nogales, Arizona, United States, is contiguous with the international border with Mexico. Its way of life—history, people, culture, and economy—is linked to its neighbors in Sonora, Mexico. Nogales, the largest city in Santa Cruz County, is largely of Hispanic (94.5%) and Mexican origin [27]. Community health workers (CHWs), who are frontline workers and have a close relationship with the community [28], are central to the MCHC health promotion efforts, including cervical and breast cancer screening, navigation services for cancer treatment, and supportive care after treatment. Breast cancer is the most common cancer type among women served by MCHC. MCHC CHWs have facilitated a breast cancer support group for >20 years in the Nogales community. The cancer support group participants developed a strong network of mutual support and provided ongoing guidance and input for the project.

Participants

The MCHC CHWs (TE and LG) currently facilitating the cancer support network engaged cancer support group members in creating the program name *Vida Plena*, derived from *vivir una*

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vida plena, meaning to live a full life. This name resonated with survivors of cancer because they maintained that life was much more than their cancer diagnosis. Vida Plena was used to identify all the study-related materials. To gain additional perspectives on survivorship experience, this study included both survivors of cancer and their caregivers.

For survivors of cancer, participants were eligible to participate in the study if they were of Hispanic origin, aged ≥18 years, had a history of breast cancer, were female, and were diagnosed with cancer in the previous 15 years. For cancer caregivers, the eligibility criteria included self-identification as a caregiver for an individual with cancer. No sex restrictions were applied. MCHC CHWs recruited participants through referral by MCHC health care providers and CHWs, and electronic flyers delivered to a pre-existing cancer support group chat through *WhatsApp*. Eligible survivors of cancer were asked if their caregivers were interested in participating.

Research Ethics Board and Informed Consent

MCHC CHWs (TE and LG) and research staff (RMV and MLP) recruited all participants and assisted with the informed consent process. Informed consent was obtained from all participants included in the study. All study materials were available in English and Spanish. Participants were reimbursed US $25 for their time. The study protocol was approved by the University of Arizona Institutional Review Board (2005660838). Study procedures were completed accordance with the ethical standards of the responsible committee on human experimentation and with the Helsinki Declaration of 1975, as revised in 2000.

Data Collection

Owing to current public health recommendations at the time of this study related to reducing the spread of SARS-CoV-2 (COVID-19) and the exposure risk for survivors of cancer [29], all study activities, including recruitment, consent, interviews, analysis, and interpretation, were completed remotely via the internet and telephone. The study was conducted entirely in Spanish as the preferred language of participants. The existing relationship of the CHW with community members was essential to our success in overcoming barriers to recruitment to this internet-based interaction.

After enrollment, a bilingual and bicultural research staff member (RMV or MLP) contacted the participants via telephone to administer a short questionnaire on demographics and diet and physical activity behaviors before and after cancer treatment, adapted from the Women's Healthy Eating and Living Study [30]. The research staff member scheduled a separate appointment for the semistructured interviews, which included 10 open-ended questions related to the impact of cancer on lifestyle, perceptions of lifestyle, and intervention content and delivery. The interviewer used probe cues to elicit deeper responses. The interviewer incorporated important cultural values that may influence lifestyle. Defined norms and roles in Mexican culture (eg, establishing warm interpersonal relationships, respect, and pleasant social exchanges) were applied during all the interviews [31]. The same interview guide was used to interview survivors and caregivers. The guide was developed with MCHC CHWs and is detailed in Table 1. All interviews were completed using Zoom software (Zoom Video Communication Inc) and were audio recorded, with permission, using integrated Zoom features for transcription and analysis. With Zoom, participants could complete the interview through the internet or telephone dial-in; video was disabled during internet-based calls to assure that all interviews relied on a consistent, audio response to queries. Each interview lasted approximately 1 hour.

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Table 1. Interview question guide for Vida Plena.

Construct	Question (English)	Question (Spanish)	Probe
Impact of cancer on lifestyle			
	What does an average day look like for you?	¿Cómo es un día normal para usted?	• Meals • Social • Family • Work • Routines • Weekend vs weekday
	What helps you live a healthy lifestyle?	¿Qué le ayuda a vivir un estilo de vida saludable?	• Facilitators
	How is your lifestyle different after cancer treatment?	¿Cómo es diferente el estilo de vida después del tratamiento del cáncer?	• Nutrition or diet • Physical activity or exercise
	What makes it difficult to live a healthy lifestyle?	¿Qué hace que sea difícil vivir un estilo de vida saludable?	• Barriers • Obstacles
	What things would make it easier for you to live a healthy lifestyle?	¿Qué le facilitaría an usted tener un estilo de vida saludable?	• Support • Resources
Perceptions of lifestyle			
	What, according to you, is a healthy lifestyle?	¿Qué significa para usted un estilo de vida saludable?	• Importance of healthy eating and physical therapy after cancer treatment • Change talk or thoughts around change
	How do you feel about diet or physical activity (lifestyle) as a way to reduce disease risk?	¿Qué piensa sobre la dieta/actividad física (estilo de vida) como una forma de reducir el riesgo de enfermedad?	• Beliefs • Attitudes • Knowledge
Intervention content and delivery			
	How would you feel about receiving information and support for making healthy lifestyle choices?	¿Cómo se sentirá al recibir información y apoyo para elegir un estilo de vida saludable?	• Preferences for information • Type of support
	What is your preference for receiving information about healthy lifestyle behaviors?	¿Cuál es su preferencia para recibir información sobre comportamientos de estilo de vida saludable?	• Written materials • Web-based • Telephonic • Face to face • Individual or group
	What are the reasons for you to choose (or not choose) to participate in a lifestyle program?	¿Cuáles son las razones por las cuales elegiría (o no elegiría) participar en un programa de estilo de vida?	• Access • Norms

Data Analysis

Descriptive statistics were completed using STATA (version 16.1; StataCorp LLC). Audio recordings of interviews were transcribed using Google Speech-to-Text application programming interface (Alphabet Inc) with quality control by native Spanish speakers by the Behavioral Measurement and Interventions Shared Resource [32] at the University of Arizona Cancer Center. Authors (MLP and MI) coded the interviews using the Quality of Cancer Survivorship Care Framework [18] and developed codebooks for survivors and caregivers in consensus with MCHC partners. The thematic framework included five domains for cancer survivorship quality: health promotion (lifestyle and cancer-preventive behaviors), recurrences and new cancers (cancer screening practices), physical effects (symptoms resulting from cancer treatment), psychosocial effects (psychosocial symptoms, financial impact, and employment), and chronic conditions (non–cancer-related conditions). Verified transcripts were independently dual coded in Spanish by MLP and MI for relative themes and content using Dedoose 8.3.43 (SocioCultural Research Consultant LLC). After initial coding of the first 2 interviews, MLP and MI met to discuss definitions and resolve any discrepancies in coding. On the basis of this discussion, the code spirituality or religion was added to capture thematic references to the participants' faith or spiritual beliefs. The remainder of the interviews were coded using the revised codebook. A directed content analysis approach was used to conceptualize a theoretical framework and highlight relationships [33].

A participatory process for thematic analysis and interpretation with the MCHC included weekly meetings and discussions of themes throughout the analytic process with the entire research team. The coded domains interrelate and are often codependent

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on one another, and thus, subthemes that evolved in collaboration with MCHC CHWs included diet, physical activity, family, support, and finance. The 2 perspectives on survivorship (survivor and caregiver) were analyzed using the same framework. The overlap of themes is shown in Figure 1.

Figure 1. Theme overlap and co-occurrence among survivors of breast cancer and caregivers participating in Vida Plena. Numbers across the top row of the table correspond to the numbered theme listed in the first column. Numbers in each cell correspond to the count of code co-occurrences among themes. Color categories correspond to the count (white=0, blue=1-10, green=11-30, yellow=31-49, and red=50).

Codes	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	Total
1. Access to care		0	0	1	0	0	0	5	4	1	1	1	2	0	1	16
2. Caregiver experience	0		1	0	0	2	0	8	2	0	3	1	2	0	1	20
3. Chronic conditions	0	1		0	1	0	0	7	2	3	0	0	2	0	1	17
4. Cultural references	1	0	0		2	3	0	4	5	0	0	1	1	1	0	18
5. Emotional wellness	0	0	1	2		6	1	10	3	5	2	16	1	5	5	57
6. Family	0	2	0	3	6		1	10	14	3	4	8	2	3	15	71
7. Finance	0	0	0	0	1	1		2	3	2	0	1	0	0	3	13
8. Health promotion	5	8	7	4	10	10	2		50	28	9	8	16	0	33	190
9. Nutrition	4	2	2	5	3	14	3	50		16	11	5	11	0	13	139
10. Physical activity	1	0	3	0	5	3	2	28	16		9	3	6	0	7	83
11. Physical effects	1	3	0	0	2	4	0	9	11	9		3	3	0	3	48
12. Psychosocial effects	1	1	0	1	16	8	1	8	5	3	3		2	5	3	57
13. Recurrences and new cancers	2	2	2	1	1	2	0	16	11	6	3	2		0	1	49
14. Spirituality or religion	0	0	0	1	5	3	0	0	0	0	0	5	0		0	14
15. Support	1	1	1	0	5	15	3	33	13	7	3	3	1	0		86
Total	16	20	17	18	57	71	13	190	139	83	48	57	49	14	86	

Results

Participants

A total of 26 individuals recruited by the CHWs were contacted for an interview (16/26, 62%, survivors; 10/26, 38%, caregivers). Of the 26 individuals, 3 (12%) were not eligible (2 survivors and 1 caregiver) and 2 (8%) caregivers dropped out after enrollment. Of the 21 participants, all of them (100%) completed the questionnaires (14 survivors and 7 caregivers) and 19 (91%) completed interviews (12 survivors and 7 caregivers).

All survivors of cancer were Spanish-speaking women, with a history of breast cancer. Most cancer caregivers were women and were daughters, mothers, or friends of the survivors. The average age of survivors of cancer was 57.4 (SD 12.4) years compared with 41.1 (SD 15.3) years for cancer caregivers. All participants were of Mexican origin, and most of the participants were Mexican-born. All survivors of cancer reported having at least one comorbid condition, with most of them having 4 or more. The most common comorbid conditions among survivors of cancer were vision problems (9/14, 64%), high cholesterol (6/14, 43%), and type 2 diabetes mellitus (4/14, 29%). The most common posttreatment symptoms included pain (6/14, 43%), sleep disorders (5/14, 36%), and depression or anxiety (4/14, 29%). Information on comorbid conditions experienced by cancer caregivers was not collected. Participant characteristics are detailed in Table 2. With regard to technology use, most survivors of cancer and caregivers used SMS text messaging and the internet several times a week or more, and all of them currently owned a smartphone (Table 3).

Many self-reported lifestyle behaviors of survivors of cancer and caregivers changed from before and after diagnosis (data not shown). After diagnosis, many survivors and caregivers reported increasing their intake of fruit (10/21, 48%), vegetables (10/21, 48%), fish (11/21, 52%), poultry (12/21, 58%), nuts (7/21, 33%), and whole grains (8/21, 38%) and decreasing their intake of red meat (14/21, 67%), fried food (14/21, 67%), fast food (8/21, 39%), and sweets (8/21, 39%) compared with before cancer diagnosis. The most commonly reported physical activities were walking (13/21, 62%) and dancing (13/21, 62%), with a low reported prevalence of mind-body physical activity (eg, Tai Chi and yoga; 2/21, 10%). Prayer was described as a form of meditation. For most participants, there was no change in physical activity after cancer diagnosis, with the exception of walking, which showed a 53% (11/21) increase.

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Table 2. Demographics and health characteristics of participants enrolled in Vida Plena (N=21).

Demographics	Survivor (n=14)	Caregiver (n=7)	Total
Age (years), mean (SD)	57.4 (12.4)	41.1 (15.3)	52.0 (15.2)
Sex (female), n (%)	14 (100)	5 (71)	19 (90)
Education, n (%)			
High school or less	7 (50)	4 (57)	11 (19)
Bachelor's degree or higher	7 (50)	3 (43)	10 (38)
Hispanic population[a], n (%)	13 (100)	7 (100)	20 (100)
Mexican origin, n (%)	14 (100)	7 (100)	21 (100)
Lived in current neighborhood (years), n (%)			
<10	5 (36)	4 (57)	9 (43)
≥10	9 (64)	3 (43)	12 (57)
Spanish primary language spoken, n (%)	14 (100)	5 (71)	19 (90)
Currently married, n (%)	8 (57)	4 (57)	12 (57)
Employment, n (%)			
None	12 (86)	3 (43)	15 (71)
Part time or full time	2 (14)	4 (37)	4 (19)
Cancer diagnosis, n (%)			
Breast	14 (100)	N/A[b]	N/A
Cancer stage at diagnosis, n (%)			
Stage 1	3 (21)	N/A	N/A
Stage 2	5 (36)	N/A	N/A
Stage 3	3 (21)	N/A	N/A
Not known	3 (21)	N/A	N/A
Age diagnosed (years), mean (SD)	49.6 (12.4)	N/A	N/A
In active treatment, n (%)	7 (50)	N/A	N/A
Family history of cancer, n (%)	10 (71)	5 (71)	15 (71)
BMI (kg/m^2), mean (SD)	28.8 (6.2)	29.7 (5.7)	29.1 (5.9)
Ever smoked, n (%)	3 (21)	3 (43)	6 (29)
Any comorbid condition, n (%)	14 (100)	N/A	N/A
Count of comorbid conditions[c], n (%)			
None	0 (0)	N/A	N/A
1-3	6 (43)	N/A	N/A
≥4	8 (57)	N/A	N/A

[a]Missing data n=1.

[b]N/A: not applicable.

[c]Comorbidities include type 2 diabetes, asthma, depression or anxiety, heart disease, vision problems, arthritis, chronic gastrointestinal conditions, sleep disorder, high cholesterol, pain, and other comorbid conditions. Comorbidity data were not collected for cancer caregivers.

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Table 3. Technology use among survivors of cancer and caregivers participating in Vida Plena (N=21).

Technology use	Survivor (n=14), n (%)	Caregiver (n=7), n (%)	Total, n (%)
Own a cell phone	14 (100)	7 (100)	21 (100)
Own a smartphone	13 (93)	7 (100)	20 (95)
How often use SMS text messaging			
Rare to never	3 (21)	1 (14)	4 (19)
Several times a week	2 (14)	2 (29)	4 (19)
At least once a day	1 (7)	1 (14)	2 (10)
Many times a day	8 (57)	3 (43)	11 (52)
How often use the internet			
Rare to never	4 (29)	1 (14)	5 (24)
A few times a month	1 (7)	0 (0)	1 (5)
Several times a week	2 (14)	1 (14)	3 (14)
At least once a day	1 (7)	1 (14)	2 (10)
Many times a day	6 (43)	4 (57)	10 (48)

Qualitative Results for Survivors

Health Promotion

Barriers Related to Healthy Eating

Many survivors of cancer reported that eating healthy was an important lifestyle habit for the prevention of cancer recurrence and the development of cancer in their family members. Survivors reported that among the barriers to healthy eating was the idea that traditional northern Mexican foods are unhealthy. Specifically, one survivor highlighted that Mexican dishes can be very greasy:

> *I believe that the biggest barrier in terms of the culture where I come from is the food that has been instilled in us, the style of food is very, very fatty sometimes, all Mexican dishes are very fatty* [Survivor #1 (original quotes in Spanish are available in Multimedia Appendix 1)]

In addition, participants alluded that eating healthy was difficult to do because of the lifestyle of the people around them. Specifically, food choices, time of meals, and food quantity were influenced by the participant's home and social environment, "for me, one of the obstacles in my life is the lifestyle of the people with whom I live" (Survivor #16).

When and What Not to Eat

In total, 2 main themes emerged among survivors of cancer related to dietary changes and the adoption of healthier dietary habits. The first identified theme specific to dietary changes among participants was the concept of eating *on time*. Survivors consistently reported a belief that eating each meal at specific times of the day was an important aspect of healthy eating:

> *If you want to live a healthy life you should eat breakfast at exactly 8 in the morning, eat at 2 in the afternoon, and have dinner at 8 at night* [Survivor #14]

Second, there was a clear focus on specific foods to avoid or consume less. For example, survivors of cancer reported focusing efforts on staying away from or reducing the intake of canned foods, sugar, dairy, meat, and soy. Multiple participants reported reducing the number of tortillas and *harinas* (flour-based products) in their diet after diagnosis:

> *I think one should eat less of everything, for example for many years I have been eating just two tortillas and that is it* [Survivor #2]

Importantly, when speaking about dietary changes for a healthier lifestyle, survivors of cancer emphasized reducing or avoiding what they considered unhealthy foods but did not report the foods or food groups they felt they could eat more of to add to a healthy diet.

Access to Reliable Diet-Related Information

Survivors reported a lack of reliable diet information as a common barrier. Survivors of cancer narrated their personal experiences in accessing dietary information through the internet and the difficulty in identifying quality and reliable information. Survivors reported the need to have access to a nutrition professional during and after their cancer treatment

> *I have always thought that people who enter cancer treatment and survivors, just like they send us to physical therapy after our surgeries, they should also send us to a nutrition class or to the nutritionist* [Survivor #1]

Participants also expressed their desire for nutrition education not only for themselves, but for their families:

> *We have to re-educate the whole family...I have yet to find resources that provide support to the family to change the eating habits, therefore it is up to me to educate them* [Survivor #1]

Role of Physical Activity After Treatment

Survivors recognized the benefits of physical activity in overall health and for the prevention of cancer recurrence. Importantly,

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a common theme was the use of physical activity as a tool to manage physical effects after cancer treatment:

> *Exercise is now more necessary for me because my arm, well, you can see that I get numb, and if I am not exercising it becomes heavy and I feel swollen...then I have to exercise to remove that feeling* [Survivor #11]

The most commonly reported physical activity was walking with dancing and biking, similar to other reported activities.

Physical Activity Gradually Decreases Over Time

A common theme among survivors of cancer was recognition that physical activity is beneficial but very difficult to maintain over time because of several factors, such as lack of motivation:

> *It has been very difficult for me lately to exercise, getting on the bike is very difficult because I feel that it is like something psychological and I am going to get tired* [Survivor #14]

Survivors of cancer also reported engaging in less physical activity as they get further away from their initial cancer diagnosis:

> *I used to exercise at the beginning, but right now I don't do it, I used to go out for a walk but now I don't* [Survivor #13]

Participants also expressed concerns regarding the safety of engaging in physical activity where they live, "It is difficult to find a safe place to walk" (Survivor #14), whereas a participant on the US side of the border had safety issues related to wildlife such as "javelinas [native wild pig-like animal] and snakes" (Survivor #10) in surrounding areas where she usually exercises.

Awareness of Cancer Etiology

Survivors mentioned in the context of cancer recurrence awareness of modifiable and nonmodifiable risk factors for cancer (ie, genetics, sex, and diet) and how this contributes to motivation for engaging in healthy lifestyle behaviors. For example, Survivor #6 particularly mentioned, "In my case, the cancer I got was genetic," whereas Survivor #1 referred to her cancer as being hormone-based, "In my case, my cancer is very hormone-influenced." Given this awareness, survivors of cancer understood not only the ongoing risk for recurrence but also the repercussions for their family members, "Family is also more likely to get cancer and that gives motivation for everyone to stay healthy" [Survivor #3]. Survivors of cancer reported on knowing the value of following recommendations and guidelines for cancer surveillance and prevention, "In my checkups that I get done every six months, thank God everything has looked good" [Survivor #16].

Survivor of Cancer Support Network

Participants reported they would like to meet with other survivors of cancer to share their experiences, from physical effects of the treatment to sharing information on other lifestyle factors such as dietary changes and recipes:

> *Being in a group where we all have the same illness, we are all going to talk, we are going to hear opinions of others...give information about how it was or what it has felt, there we see that we are not all the same* [Survivor #13]

Similarly, another noted, "What motivated me a lot were the cancer survivor classes I attended, I liked it a lot," (Survivor #5) highlighting how other survivors of cancer are indeed a source of support and motivation.

Psychosocial and Physical Effects

Physical and Emotional Support

Survivors of cancer reported that the primary way they received support from family was through acts of service. Survivors reported that delegating responsibilities to their partners and children was imperative while navigating their cancer treatment:

> *During my second treatment, I was bed-bound and felt sicker, therefore, I learned I had to delegate responsibilities to my daughters, who have always been there, and my husband* [Survivor #10]

These included responsibilities within the household (ie, cooking and cleaning):

> *My children come and cook for us, often we have a barbecue and we spend time with them...my husband takes care of the grocery shopping* [Survivor #5]

Survivors also expressed that family members played a significant role in providing emotional support and motivation:

> *My children and grandchildren with me motivation to do what I have to do even when I am not in the mood to do it* [Survivor #10]

Family Support for a Healthy Lifestyle

Participants reported that family provided a significant amount of support in engaging in health-promoting behaviors. Several survivors of cancer expressed that when it came to physical activity, family members would also engage in physical activity with them:

> *My husband and my son are the ones that go with me on walks, my son runs with me as well* [Survivor #2]

Oftentimes, the support of family in physical activity was crucial for the survivor of cancer to engage in physical activity:

> *Once my husband gets home from work, we go for a walk outside for like a mile or a mile and a half* [Survivor #2]

Support for healthier eating habits was different from that for physical activity. Many survivors of cancer reported feeling less support from family members when it was related to adopting healthier eating habits than physical activity:

> *When you are around your family or other people, what they eat is what you are going to have to eat* [Survivor #1]

Oftentimes, survivors reported that family members have different food preferences that may not align with their goal of adopting healthy dietary habits and that it was difficult to cater to everyone in the family:

> *My husband likes fried foods, he loves all those fried foods, therefore, cooking foods for him or for someone*

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else in the house based on their preferences is a barrier because you can't cook three different dishes at the same time [Survivor #1]

Importance of Spirituality or Religion

Spirituality or religion provided survivors both motivation to live a healthy life and as a way to cope with their cancer diagnosis:

I do know that it is very important that my God thinks it was worth to let me live one more year, and to think it is worth letting me live another one so I constantly evaluate myself [Survivor #6]

Participants often had statements or interjections nested within other themes with a contextual religious lens through which they were looking at their experiences such as "thank God" (Survivor #2) or "God willing" (Survivor #10).

Cancer Treatment-Related Symptoms Interfere With Lifestyle

Lymphedema was the most commonly reported physical effect during the interviews among survivors. Participants often reported the ongoing struggle of dealing with lymphedema in activities of daily living and influencing ability to engage in physical activity. One survivor mentioned:

There are things that, for example, I can no longer lift, things that I used to lift heavy or things like that...now I'm trying to regain that mobility in my life but I know that maybe I won't get it back 100% due to lymphedema [Survivor #1]

The connection between physical activity and minimizing the effects of lymphedema was also mentioned by the participants. There were no other mentions of coexisting chronic conditions.

Financial Barriers to Live a Healthy Lifestyle

Survivors of cancer reported that financial difficulties were a major barrier to engaging in a healthy lifestyle:

Having more economic help would help me relax given I am a single person...I have been working for myself and my own wellbeing [Survivor #14]

With regard to diet, many participants held the belief that eating healthy is more expensive and identified lack of finances as a barrier for acquiring healthful foods:

We know that leading a healthy lifestyle is always a little more expensive because we have to buy vegetables [Survivor #16]

Similarly, another participant reported that oftentimes fast food can be cheaper:

Many times the food that we have to eat or have to prepare is more expensive than buying junk food [Survivor #3]

Likewise, financial barriers were reported to engage in physical activity. Participants reported that financial difficulties prevented them from acquiring or accessing equipment or spaces for physical activity, "It has been a while since we are trying to buy a treadmill and I still can't buy it" (Survivor #2) and "Having money to buy what we have to in order to lead a healthy lifestyle...like going to the gym" (Survivor #16).

Qualitative Results for Caregivers

Caregiving Experience Effect on Lifestyle Habits

Caregivers reported that seeing survivors of cancer navigating cancer treatment increased their awareness and influenced them to engage in healthier lifestyle behaviors themselves. For example, participants reported increasing physical activity levels and making additional efforts to eat healthy with the purpose of "keeping the cancer away" (Caregiver #3) not only for themselves but also within their families: "If we all eat healthy and exercise, the cancer won't be back in our family" (Caregiver #3) (original quotes in Spanish are available in Multimedia Appendix 2). Smoking cessation was acknowledged as an important aspect in maintaining a healthy lifestyle and one caregiver reported quitting smoking after their loved one was diagnosed:

Me for example, before the cancer treatment, I used to smoke cigarettes, but from the moment of diagnosis, that is when I changed this habit because I realized I was hurting myself. [Caregiver #3]

Cancer diagnosis also had a positive effect on other cancer prevention screening measures among caregivers. For one caregiver in their family, being proactive and engaging in precautionary measures for cancer prevention was a new adopted habit:

Before I did not check myself and now I go once a year, I always go and have my ultrasound, my mammogram, or I go with them [sisters who are survivors of cancer] when they have a check-up and yes, a disease like this affecting your family it really changes your life. [Caregiver #10]

Although the cancer experience of a loved one was reported to be a source of motivation for improving their own lifestyle, caregivers also reported a decreased effect over time. For example, Caregiver #3 mentioned that although they were caring for the survivor of cancer, they were more diligent in engaging in healthy lifestyle habits, whereas now, the fact that survivors of cancer and caregivers have their own separate routines has diminished this effect.

Supporting the Survivor

Providing support for survivors, particularly by taking care of activities related to their daily living, was commonly reported. Daily routines changed during their loved ones' cancer treatment to provide support for activities of daily living and emotional needs to manage side effects (ie, hair loss, lack of appetite, or mood). Oftentimes, there was a balance between responsibilities and time:

When I was taking care of my mother, I would get up much earlier than normal to be able to take care of my responsibilities; go to drop off the children at school and come back so that I could help [my mom] clean her house, make her breakfast, and when she had the surgery, I would change her dressings [Caregiver #3]

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One caregiver reported it is crucial to:

> *give them a lot of affection so that they do not feel alone, that all things happen and that this will definitely happen but above all give them a lot of love, a lot of trust, a lot of assurance that they are not alone* [Caregiver #4]

Importantly, caregivers highlighted their experience and caregiving duties to be an honor rather than a burden, one considered it, "gratifying for oneself" and highlighted that:

> *We do not know if tomorrow one may be in the same situation and I want to think that there will always be someone you can count on* [Caregiver #8]

Discussion

Principal Findings

This is the first qualitative study to explore Mexican-origin survivors of breast cancer and caregivers' perspectives on lifestyle behaviors, namely, diet and physical activity, and important aspects for developing and adapting a culturally relevant lifestyle intervention for survivors of cancer residing in border communities. Using a community-based participatory approach between an academic partner and a community partner enabled us to recruit and conduct interviews with 12 survivors of breast cancer and 7 caregivers. The perspectives of bilingual and bicultural members of our team allowed us to identify relevant and culturally specific perspectives related to the impact of cancer on lifestyle, perceptions of lifestyle, and intervention content and delivery preferences. Key themes, which emerged through framework guided directed thematic analysis, included health promotion and psychosocial and physical effects among survivors of cancer, whereas only health promotion was a key theme among caregivers. Family was a central and recurring subtheme throughout as was the desire for access to content experts for health promotion education.

Participants expressed an awareness of the role of nutrition and physical activity in cancer prevention and survivorship. Physical activity was linked with physical effects (where physical activity improved side effects related to cancer treatment and side effects interrupted abilities to engage in physical activity) and was considered more challenging to engage in compared with other recommended lifestyle habits (eg, diet). Family provided physical and emotional support, and healthy lifestyle behaviors (or lack thereof) were experienced as a family unit. Variations were noted among caregivers, including that the cancer diagnosis was influential to their lifestyle behaviors and that there was pride in supporting the survivor. Unique to this study was the self-reported diet and physical activity before and after treatment in both the survivor and caregivers, which provided quantitative support for our qualitative findings related to lifestyle behaviors.

Our findings revealed an emphasis on what not to eat and a reported lack of reliable information. Diet plays an important role in Mexican culture, with traditions and social components centered on planning, cooking, and eating foods [34]. Participants in this study reported eating typical Mexican foods with family and friends as a common quality time activity with loved ones. However, Mexican food was commonly perceived as unhealthy and was labeled as a barrier for healthy eating because of its *very greasy* nature. Concordant findings were reported by Ramirez et al [35], where participants reported that "growing up in a Mexican household, a lot of the foods that [they] eat aren't very healthy." Additional findings from the study by Ramirez et al [35] described how typical Mexican foods (ie, pozole and tamales) are perceived as foods eaten as a *treat* and are not foods that constitute a healthy eating pattern. Interestingly, several reports in the literature have focused on describing and investigating a *traditional Mexican diet* and its effects on health [36-40], which contrast with the reported belief of the overall Mexican diet being unhealthy, as observed in our study and others. Overall, a traditional Mexican diet is characterized by higher amounts of plant-based foods, including, but not limited to, legumes, grains, and vegetables and high amount of specific foods such as maíz (corn), beans, chile, squash, onion, and garlic [41]. A traditional Mexican dietary pattern has been associated with lower systemic inflammation, lower insulin sensitivity, lower risk for overweight and obesity, and lower risk of obesity-related cancer mortality collectively across several studies, including a randomized controlled feeding trial and other epidemiological studies [36-40]. Importantly, the participants in our study reported several strategies that would help them overcome reported barriers to healthy eating. Among the top requests, having access to an informed nutrition professional and participating in survivorship support groups were highly requested. Such strategies should be considered as future dietary intervention components among border-dwelling Mexican-origin survivors of cancer.

The Quality of Cancer Survivorship Care Framework [18] used to analyze our interviews can be applied to intervention design as well to include tailored risk assessment for behaviors, symptoms, finances, and interpersonal relationships in addition to education provided by clinical professionals and care coordination. Our results further emphasized the need for reliable information related to diet and physical activity and the importance of content knowledge experts, such as registered dietitians, to be involved in intervention planning and delivery. Engagement of culturally competent knowledge experts throughout intervention design and delivery is a way to provide reliable education and increase participants' acceptability and access to clinical professionals. As the experience of physical effects and comorbidities is heterogeneous in survivorship, access to clinical professionals may be required to provide tailored advice on how to safely engage in physical activity and dietary change. Important considerations for intervention planning include the perceived costs observed in this study related to engaging and maintaining a healthy lifestyle and feelings of safety around physical activity.

Highlighted by the intersection of the survivor, their family connections, and desire for community and structural support for a healthy lifestyle, delivery of an intervention could further benefit from theoretical guidance from the socioecological model. Theoretical constructs are associated with meeting current diet and physical activity recommendations among survivors of cancer, and theory-informed interventions often have a sustained impact on behavior change in survivors of cancer [42,43]. The socioecological model describes the

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complex interactions between individuals nested within relationships and environments. Successful interventions built from the socioecological model include components of education, skills enhancement, modifications to home or institutional environments, community capacity, and policy advocacy [44] and inclusion of the survivorship community (including survivors of cancer, caregivers, providers, advocates, public health professionals, and policy makers) [45]. The socioecological model may promote better outcomes for survivors of cancer [45].

Strengths and Limitations

Strengths of this study include the value of a participatory approach. The effective collaboration between the investigating team and community partners allowed for innovative and successful completion of the study, particularly in a highly unpredictable time such as early during the COVID-19 pandemic. This approach allowed the development of strategies that were feasible for implementation among study participants, such as conducting and completing all study-related activities remotely and troubleshooting technologic difficulties that resulted in little to no issues with participants accessing the videoconferencing software and further expanded our reach to rural survivors of cancer. This study was limited by the breast cancer perspective only and may not be translated to all obesity-related cancers or the cancer experience in male survivors of cancer. In addition, caregiver experiences may be limited in their generalizability to other cancer populations. The Quality of Cancer Survivorship Care Framework [18] used to guide our analysis did not translate seamlessly into caregiver interviews, and some of the themes from the framework were minimal in our results. This warrants for the use of a caregiver specific framework, such as the one developed by Fletcher et al [46], which seeks to incorporate relevant factors for this population such as culture, socioeconomic status, and access to health care when analyzing data as the one presented in this qualitative study.

Recommendations for Intervention Planning and Next Steps

In summary, the future development of culturally appropriate and acceptable lifestyle interventions to improve cancer survivorship among Hispanic populations living in the United States–Mexico border may consider the following points:

1. Culturally relevant and competent content consisting of traditional Mexican diet and social norms
2. Assess and address barriers, including physical and psychological side effects of cancer treatment and environmental and financial factors
3. Multimodal programming composed of written educational materials, interactive support groups, and nutrition or physical activity content experts
4. Inclusion of a survivor-identified informal caregiver or family members
5. Implementation strategies to investigate integration into a clinical setting with delivery by CHWs

Acknowledging the heterogeneity of the Hispanic population, studies should focus on programs that target populations from different countries of origin and regions of the United States separately to address the specific lifestyle behavior needs and barriers to the target population and evaluate which cultural adaptations are well received and effective in improving feasibility, acceptability, and replication of culturally tailored programs. Given that different cancer sites and treatments may yield distinct complications and side effects, cancer diagnosis, treatment, and subsequent treatment-related side effects influence on lifestyle behaviors should be considered. Forming or continuing community partnerships and collaboration can establish effective and sustainable lifestyle behavior programming for advancing cancer survivorship among Hispanic populations.

Conclusions

Our study identified that Mexican-origin survivors of breast cancer desire relevant and evidence-based information related to healthy lifestyle behaviors and highlight the influence of family and community on the adoption of a healthy diet and physical activity habits. These qualitative findings and recommendations support a theory-informed, evidenced-based, CHW-led, culturally relevant lifestyle program to reduce the burden of cancer recurrence, comorbidities, and potential outcomes after cancer in Mexican-origin survivors of cancer living in border communities and cancer prevention among cancer caregivers.

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Authors' Contributions

All authors contributed to the conception and design of the study. Methods and material preparation were carried out by MBS, RMV, MI, TE, LG, and CAT; data collection was carried out by RMV, and MLP; data analysis was carried out by MLP, MI, MBS, and SJW; data interpretation was carried out by MBS, MLP, SJW, MI, TE, LG, RMV, and CAT; and supervision and

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funding acquisition was carried out by CAT. The first draft of the manuscript was written by MBS and MLP, and all authors commented on the previous versions of the manuscript. All authors read and approved the final manuscript.

Conflicts of Interest

None declared.

Multimedia Appendix 1
Identified themes and quotes among survivors of cancer (n=12).
[DOCX File , 103 KB - cancer_v8i1e33083_app1.docx]

Multimedia Appendix 2
Identified themes and quotes among caregivers (n=7).
[DOCX File , 102 KB - cancer_v8i1e33083_app2.docx]

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Abbreviations

CHW: community health worker
MCHC: Mariposa Community Health Center

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Original Paper

Evaluation of the Pathways for Survivors Program to Address Breast Cancer Survivorship–Associated Distress: Survey Study

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Abstract

Background: Patients with breast cancer frequently experience escalation of anxiety after completing curative treatment.

Objective: This study evaluated the acceptability and psychological impact of a 1-day workshop to emphasize behavioral strategies involving intention and self-efficacy.

Methods: Breast cancer survivors who attended a 1-day Pathways for Survivors workshop provided feedback and completed electronic quality of life (QOL) questionnaires at baseline, 1 and 6 weeks, and 6 months after the workshop. Attendees' baseline QOL scores were compared to follow-up (FUP) scores. Scores from patients receiving routine FUP care were also compiled as a reference population.

Results: In total, 77 patients attended 1 of 9 workshops. The mean satisfaction score was 9.7 out of 10 for the workshop and 9.96 out of 10 for the moderator. Participants' baseline mean Patient-Reported Outcomes Measurement Information System (PROMIS) anxiety and depression scores were 57.8 (SD 6.9) and 55.3 (SD 7.5), respectively, which were significantly higher than those of patients receiving routine FUP care (49.1, SD 8.3 and 47.3 SD 8.0, respectively). PROMIS anxiety and depression scores decreased, and the Happiness Index Profile (HIP-10) score—measuring intention and resiliency—increased significantly at 1- and 6-week FUPs.

Conclusions: The Pathways for Survivors program was favorably received. Anxiety and depression decreased significantly at 1- and 6-weeks after the workshop and remained below baseline at 6 months. Increased HIP-10 scores suggest that patients acquired and implemented skills from the workshop. A 1-day workshop led by a lay moderator significantly improved several psychological measures, suggesting that it may be a useful and time-efficient strategy to improve QOL in breast cancer survivors. We are investigating whether an abbreviated "booster" of the intervention at a later date could further improve and maintain QOL gains.

(JMIR Cancer 2022;8(1):e31756) doi:10.2196/31756

KEYWORDS

breast cancer; depression; anxiety; quality of life; breast cancer survivors; cancer survivorship; mental health; psychological health

Introduction

There were an estimated 3.8 million breast cancer survivors in the United States in 2019, and this number is expected to be close to 5 million by 2030 [1]. Transitioning from a patient with cancer to a cancer survivor is challenging, and many patients with breast cancer have unmet physical and emotional needs [2-5]. Studies have found increased rates of anxiety and

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depression among breast cancer survivors over the short and long term, and these problems appear more prominent in younger survivors and those with pre-existing psychological symptoms [4,6,7]. Many studies also identify fear of cancer recurrence (FCR) and difficulty in returning to "normalcy" as potential sources of distress in this population [4,8,9]. The nature of intrusive thoughts associated FCR have been shown to share many characteristics with worry or anxiety [10]. Moreover, a systematic review of adult cancer survivors found that depression and anxiety were significantly correlated with FCR, and psychological distress is a strong predictor of FCR [11]. During the acute phase of care when attending regular medical appointments, patients often feel more secure that there is active monitoring for signs and symptoms of cancer recurrence. After active treatment ends, patients with breast cancer may feel a loss of a safety net. A comprehensive review of breast cancer survivors (≥1 year from diagnosis) showed compelling evidence of an increased risk of anxiety, depression, suicide, and neurocognitive and sexual dysfunction in breast cancer survivors compared with women with no prior cancer [6]. These findings indicate the need for novel interventions to help manage these psychological symptoms in breast cancer survivors.

The Pathways for Survivors program was developed through a collaboration between the moderator (GH) and clinicians (MM, HR, DH, and LE) at the University of California San Francisco (UCSF). The basic principles and content of the Pathways workshop are based on a positive psychology model of cognitive behavioral therapy (CBT). This model, consistent with the Broaden-and-Build theory of positive psychology, suggests that experiencing positive emotions broadens a person's awareness and encourages varied and novel thoughts and actions, which, in turn, strengthens the individual's personal skills and resources [12]. Multiple CBT interventions have been shown to decrease anxiety and depression in various breast cancer populations [13-17].

The Pathways for Survivors program teaches specific techniques for increasing positive emotions on a daily basis, equipping patients with a variety of skills and tools to improve their quality of life (QOL) in the context of life-limiting illness. The intervention is based on a system of 9 behaviors that have been shown in other contexts to enhance QOL and emotional well-being [18,19]. With the aid of grant and philanthropic funding, the UCSF Breast Care Center (BCC) has offered Pathways workshops several times a year since 2015 as a free resource to breast cancer survivors, with a focus on patients who recently completed active treatment. Qualitative and quantitative feedback on the acceptability and utility of the Pathways for Survivors program has been collected for quality improvement purposes, allowing us to better characterize the acceptability and psychological impact of the workshop. We hypothesized that this day-long workshop would have favorable effects on patient QOL by reducing both short- and long-term anxiety and depression.

This study aimed to assess the impact of the Pathways for Survivors program, a 1-day layperson-led workshop for breast cancer survivors, on breast cancer survivors' psychological distress as evaluated by a number of standardized measures of anxiety and depression. We characterized how patients received the intervention and evaluated the change in measures of patients' psychological distress from before to after the intervention.

Methods

Ethics Approval

Approval was obtained from the UCSF ethics committee (15-17099). The procedures used in this study adhere to the tenets of the Declaration of Helsinki. Informed consent was obtained from all individual participants included in the study. Patients provided written informed consent for publishing their deidentified data.

Methods Overview

Patients with stage 0-3 breast cancer, who had at least one clinic visit at the UCSF BCC and had completed their acute phase of care, including chemotherapy and breast surgery, were considered eligible and were invited to attend a day-long Pathways for Survivors workshop. Patients were recruited by their medical oncologist or breast surgeon through flyers posted in UCSF clinics, and at local breast cancer survivorship and supportive care events. For the last 2 sessions, workshops were limited to patients aged 50 years and under since the philanthropic funding to support these two workshops was intended to focus on "younger" breast cancer survivors. The workshops were conducted on a weekend day, lasting from approximately 8:30 AM to 4 PM with a 45-minute lunch break during which patients were encouraged to engage in informal interaction. The workshops were moderated by author GH and included a series of 9 lessons or exercises, most of which required substantial interaction among the participants. The central framework of the workshop was centered on "intention," which is defined in this program as "making a conscious choice toward the most beneficial thought, feeling, or behavior." Other exercises were based on the concepts of truth, accountability, identification, centrality, recasting, options, appreciation, and giving. Upon completion of the workshop, participants were asked to complete anonymous feedback surveys on program content and moderator quality.

Attendees were asked to complete a series of electronic surveys via the REDCap system at baseline (before the day-long program), 1 week after the workshop, and 6 weeks after the workshop. For the last 4 workshops, a 6-month follow-up (FUP) survey was added. Within the questionnaire, patients were presented with a consent section to have their data used for research purposes. However, patients could opt to participate in the workshop and opt out of data-sharing. Specific survey measures included the National Cancer Institute's PROMIS (Patient-Reported Outcomes Measurement Information System) anxiety and depression short-form questionnaire and the Happiness Index Profile (HIP-10) scale—a measure of psychological intention and resiliency.

The PROMIS anxiety and depression scales are two independent short-form, 4-item questionnaires that assess self-reported anxiety and depression in the past 7 days. Each item is scored from 1 (never) to 5 (always), with higher scores indicating greater anxiety or depression. PROMIS instruments were graded

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with item-level calibrations using the Health Measures Scoring Service [20] to determine PROMIS anxiety and depression T-scores.

The HIP-10 (previously HI/P6 scale) is a 10-item questionnaire assessing positive affect, intention, and resiliency. Each item is scored from 0 (strongly disagree) to 10 (strongly agree). HIP-10 scores are calculated by adding the scores for each item to generate a total score out of 100, and an increase in the score suggests greater uptake of the "intention" model. Through an independent, unpublished, pilot validation analysis in a population including college students, employees of large corporations, and retirees, the HIP-10 was found to have high internal consistency (Cronbach α=.847) and correlation with the POMS (Profiles of Moods) total scale and multiple subscales.

Within the UCSF BCC, all new patients are asked to complete an intake survey that includes demographic information, health history, and QOL instruments including PROMIS anxiety and depression. We have also implemented electronic delivery of follow-up surveys to early-stage patients in ongoing routine care. A subset of these patients agreed to have their survey data used for research. To better contextualize the Pathways patients' baseline scores within a broader general population of early-stage FUP patients at the UCSF BCC, we utilized data from patients who had completed an FUP survey, did not attend Pathways, and consented to have their data used for research. Hereinafter, these patients are referred to as the "comparison group."

The primary goal of this study was to evaluate longitudinal change in patient-reported psychological distress measures, including PROMIS depression and anxiety and HIP-10, and to evaluate demographic and clinical covariates within this population, which may help predict patients who would benefit most from this intervention.

We also compared baseline PROMIS anxiety and depression scores as well as demographic and clinical descriptors of the Pathways patients to a comparison group of early-stage FUP patients along with their PROMIS anxiety and depression scores collected at a single FUP survey.

Statistical Analysis

Descriptive statistics were used to summarize demographic and clinical data including age, stage, hormone receptor and HER2 status, nodal status, and time from diagnosis to completion of the baseline survey for Pathways participants. An independent samples *t* test and chi-square tests were conducted to compare demographics for Pathways participants and the comparison group. Independent 2-sample *t* tests were also used to compare the one-time scores on the PROMIS anxiety and depression scales of the comparison group to baseline scores of Pathways participants.

For Pathways participants, paired samples *t* tests were used to compare the PROMIS anxiety, PROMIS depression, and HIP-10 scores between baseline and the 1-week, 6-week, and 6-month scores for significance. Two-tailed *P* values of <.05 were considered significant. Among Pathways participants, analyses were conducted using paired samples *t* tests to determine if factors including age, stage, nodal status, hormone receptor status, and time from diagnosis were associated with the change from baseline in PROMIS and HIP-10 scores at 1 week, 6 weeks, and 6 months. All *t* tests used in this study are 2-tailed.

Availability of Data and Material

The data sets generated and analyzed during this study are available from the corresponding author on reasonable request.

Results

Nine sessions were held between September 2015 and December 2019. In total, 79 patients participated in the Pathways workshop and provided feedback on their satisfaction with the day-long session. A total of 77 patients consented to have their QOL data (including PROMIS and HIP-10 scores) used for research. Overall, 71 patients completed at least 1 FUP survey, of whom, 68 completed the 1-week FUP (completion rate=88%) and 61 completed the 6-week FUP (completion rate=80%). The 6-month FUP survey was sent to participants from the last 4 workshops. Of the 50 patients invited to complete the 6-month survey, 32 completed it (completion rate=65%).

Demographic data for Pathways participants and the routine follow-up comparison group patients who agreed to use of their clinically collected data for research are presented in Table 1. Pathways participants were younger than the routine FUP care patients (mean age 51.3 vs 58.5 years, *P*<.001). There were no significant differences in stage, hormone receptor and HER2 status, or nodal status. Pathways participants were, on average, 1.5 years from their diagnosis. The majority of participants were White (75.3%), well-educated (college graduates or above, 92%), and employed (45% full-time and 22% part-time).

Pathways participants had significantly higher baseline PROMIS anxiety and depression scores than the scores from a single FUP time point in the routine FUP comparison group. The baseline PROMIS anxiety mean score was 57.8 (SD 6.9) for Pathways patients versus 49.1 (SD 8.3) for the comparison group patients. Similarly, the baseline PROMIS depression mean score was 55.3 (SD 7.5) for Pathways patients versus 47.3 (SD 8) for the comparison group patients (*P*<.001 for both comparisons).

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Table 1. Demographic and clinical characteristics of Pathways participants (N=77) and early-stage routine follow-up care patients (N=71).

Characteristics	Pathways participants	Routine follow-up care patients	*P* value
Age (years), mean (SD; median)	51.4 (10.74; 51.3)	58.5 (11.79; 59.0)	<.001
Stage, n (%)			.30
0 or 1	31 (40.3)	23 (32.4)	
2 or 3	46 (59.7)	48 (67.6)	
Hormone receptor status, n (%)			.13
Negative	13 (16.9)	6 (8.5)	
Positive	64 (83.1)	65 (91.5)	
HER2 status, n (%)			.30
Negative	61 (79.2)	51 (71.8)	
Positive	16 (20.8)	20 (28.2)	
Nodal involvement, n (%)			.06
No	50 (64.9)	36 (50.7)	
Yes	26 (33.8)	35 (49.3)	
Treatment length, n (%)			N/A[a]
<6 months	22 (28.6)	—[b]	
≥6 months	55 (71.4)	—	
Race, n (%)			N/A
White	58 (75.3)	—	
Asian	11 (14.3)	—	
Other	6 (8)	—	
Not reported	2 (2.6)	—	
Education, n (%)			N/A
Some high school or less	0 (0)	—	
High school graduate or graduate equivalency degree	1 (1.3)	—	
Some college or technical school	5 (6.5)	—	
College graduate	26 (33.8)	—	
Some graduate school	2 (2.6)	—	
Master's degree	30 (39.0)	—	
PhD, MD, JD, or other	13 (16.9)	—	
Employment status			N/A
Full-time (≥35 hours/week)	35 (45.5)	—	
Part-time (<35 hours/week)	17 (22.1)	—	
Full-time parenting or caregiving	4 (5.2)	—	
Student	1 (1.3)	—	
Retired	8 (10.4)	—	
On leave/disability	7 (9.1)	—	
Other	5 (6.5)	—	
Annual income (US $), n (%)			N/A
<25,000	8 (10.5)	—	
25,000-49,999	4 (5.3)	—	
50,000-74,999	10 (13.2)	—	

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Characteristics	Pathways participants	Routine follow-up care patients	*P* value
75,000-99,999	11 (14.5)	—	
>100,000	43 (56.6)	—	
Health insurance, n (%)			N/A
Health insurance through employer	42 (54.5)	—	
Health insurance through partner's employer	18 (23.4)	—	
Private health insurance	11 (14.3)	—	
Medi-Cal/Medicare/Medicaid or some other public coverage	6 (7.8)	—	
Marital status			N/A
Married	46 (59.7)	—	
In a committed relationship	11 (14.3)	—	
Single	12 (15.6)	—	
Divorced/separated	8 (10.4)	—	

[a]N/A: not applicable.

[b]—: not available.

The distribution of PROMIS depression and anxiety and HIP-10 scores over time are depicted in Figure 1. PROMIS anxiety scores decreased significantly at 1 week (mean difference 3.884, SD 6.616; *P*<.001) and 6 weeks (mean difference 2.234, SD 7.291; *P*=.02) and showed a nonsignificant decrease at 6-months FUP (mean difference 2.466, SD 7.613; *P*=.07). PROMIS depression scores decreased significantly at 1-week (mean difference 4.260, SD 6.811; *P*<.001) and 6-weeks (mean difference 3.175, SD 6.669; *P*<.001) but increased nearly back to baseline at 6 months (mean difference 0.822, SD 6.962; *P*=.50). HIP-10 scores increased significantly at 1 week (mean difference 6.63, SD 12.41; *P*<.001) and 6 weeks (mean difference 6.21, SD 13.37; *P*<.001) and maintained a trend toward an increase at 6 months (mean difference –3.62, SD 12.811; *P*=.12). Table 2 summarizes changes in the scores for Pathways participants relative to their baseline scores.

There were no significant differences in changes in PROMIS anxiety, PROMIS depression, or HIP-10 scores of participants based on time from completion of active treatment to the time of the workshop (≤6 months vs >6 months), stage (stage 0 or 1 vs stage 2 or 3), hormone receptor status (positive vs negative) or nodal status at any follow-up point. Participants with HER2-positive breast cancer displayed a greater decrease in PROMIS depression scores than HER2-negative participants at all FUPs, although the difference was only significant at the 6-week FUP (*P*=.02). On comparing HER2-positive vs -negative participants, there were no significant differences in changes in PROMIS anxiety or HIP-10 scores from baseline to any FUP. Participants who had a shorter treatment duration (≤6 months) displayed a greater decrease in PROMIS anxiety scores than those with a longer treatment duration (>6 months) at all FUPs, although the difference was only significant at the 6-week follow-up (*P*=.049). There were no significant differences in PROMIS depression or HIP-10 scores at any FUP based on treatment length.

The average scores for satisfaction with the workshop and the moderator were 9.70 and 9.96 respectively. 98.5% would recommend the workshop to other survivors. In the immediate feedback provided at the end of the workshop, comments were all favorable and included statements such as: "This program truly gives me a pathway and an orientation of self-care. Instead of being stuck in fear, I have now a way towards a full life" and "The program offers an opportunity for "pause" in a time of great stress caused by dealing with disease and how it upends life…The skills/tools are useful in all aspects of life."

In responding to the question of what were the most helpful parts of the program, comments included the following:

> *The constant participation of everyone in the group. It was great to learn from others' experiences. The intentions and appreciations parts were my favorites.*
>
> *I most enjoyed the recasting, as it provided an intimate listening and sharing setting. I also enjoyed the appreciation line – although it was difficult, it was amazing to see connections had formed in a short space of time.*

Some comments regarding areas for improvement were the following:

> *Would be willing to do two days and/or reconnecting or having a checking in in 3 months/6 months.*
>
> *A longer program so as to allow the participants more time to share.*
>
> *Perhaps it could be done in two shorter sessions (3-4 hours each) to give the participants time to reflect on the first session before doing the second-- it's a lot to take in!*

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Figure 1. Distribution of (A) PROMIS depression T-scores, (B) PROMIS anxiety T-scores, and (C) HIP-10 scores at baseline (participants and comparison group) and follow up (participants only). BS: baseline; CNTRL: baseline comparison group; HIP-10: Happiness Index Profile; Nobs: number of observations; PROMIS: Patient-Reported Outcomes Measurement Information System.

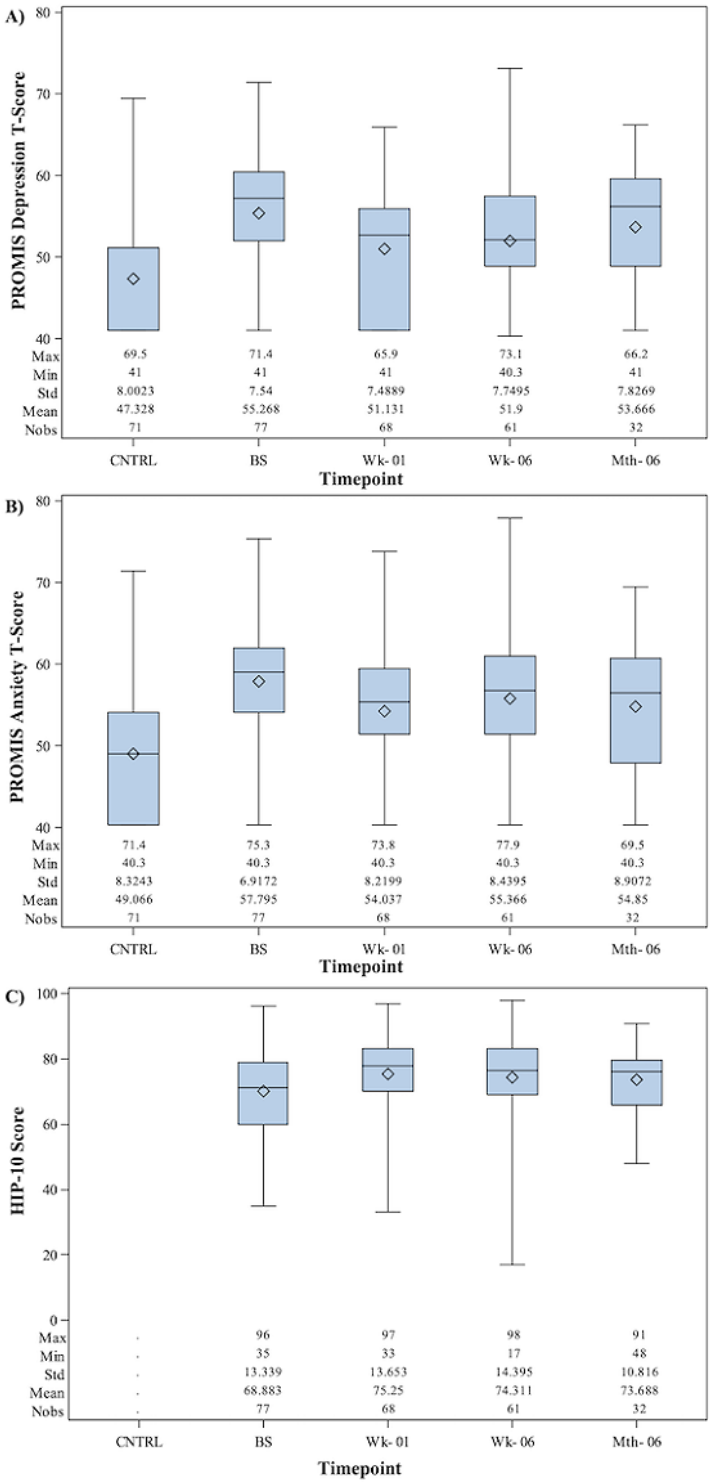

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Table 2. Summary of outcomes among Pathways participants relative to baseline.

Item	Participants, n	Score change		SE	*t* test (*df*)	*P* value
		Mean (SD)	95% CI			
Patient-Reported Outcomes Measurement Information System anxiety T-score						
Baseline to 1-week follow-up	68	3.884 (6.616)	2.282 to 5.485	0.802	4.8409 (67)	<.001
Baseline to 6-week follow-up	61	2.234 (7.291)	0.367 to 4.102	0.934	2.3936 (60)	.02
Baseline to 6-month follow-up	32	2.466 (7.613)	–0.279 to 5.210	1.346	1.8321 (31)	.08
Patient-Reported Outcomes Measurement Information System depression T-score						
Baseline to 1-week follow-up	68	4.260 (6.811)	2.612 to 5.909	0.826	5.1583 (67)	<.001
Baseline to 6-week follow-up	61	3.175 (6.669)	1.467 to 4.883	0.854	3.7188 (60)	<.001
Baseline to 6-month follow-up	32	0.822 (6.962)	–1.688 to 3.332	1.231	0.6678 (31)	.51
Happiness Index Profile score						
Baseline to 1-week follow-up	68	–6.632 (12.410)	–9.636 to –3.629	1.505	–4.4072 (67)	<.001
Baseline to 6-week follow-up	61	–6.213 (13.375)	–9.639 to –2.787	1.713	–3.6280 (60)	<.001
Baseline to 6-month follow-up	32	–3.625 (12.811)	–8.244 to 0.994	2.265	–1.6007 (31)	.12

Discussion

Principal Findings

The Pathways for Survivors workshop was well received by patients, and the overwhelming majority would recommend the workshop to other cancer survivors. Participants' PROMIS anxiety and depression scores decreased significantly up to 6 weeks after the workshop. Improvements in these QOL measures did not appear to differ on the basis of stage, time from the end of active treatment, nodal status, or hormone receptor status. Increased HIP-10 scores at 1-week and 6-week FUPs suggested that patients incorporated the intention and resiliency skills that were the focus of the workshop. While the 6-month FUPs for anxiety and HIP-10 showed a trend toward improvement compared to baseline, these results were not significant, probably owing to the smaller sample size, given that the 6-month FPU survey was only distributed to participants in the last 4 workshops, and a lower percentage (65%) of participants completed the 6-month FUP as compared to the 1- and 6-week FUPs (88 and 80%, respectively). It is also possible that the skills learned in the workshops may need to be reinforced with additional "booster" sessions. A randomized clinical trial of 8 weeks of CBT followed by 3 booster sessions in patients with metastatic breast cancer found sustained reductions in depressive symptoms and anxiety out to 6 months, which supports this hypothesis [21].

Patients who participated in the Pathways workshops, on average, had more anxiety and depression at baseline than a reference population of early-stage patients receiving routine FUP care at the UCSF BCC. Pathways participants were younger, closer to their diagnosis of breast cancer, and had more recently entered the "survivorship" phase of care than the reference group of routine FUP care patients. Notably, many of the Pathways patients were recruited by their medical or surgical oncologist to attend the Pathways workshop, and the providers likely identified patients who they thought had more psychological distress and would benefit from the intervention. Finally, the last 2 workshops were specifically targeted at younger women (<50 years of age), where the additional stresses of having children or returning to the workforce after a cancer diagnosis may be associated with greater anxiety or depression [22,23].

While it is possible that the improvements seen in the Pathways participants over time represents a natural trend of emotional and psychological recovery from the diagnosis of breast cancer and its treatment, the significant decrease in PROMIS anxiety and depression scores and improvement in the HIP-10 scores immediately after the workshop as early as the 1-week time point and sustained until 6 weeks suggests an immediate impact from the workshop. Although the intervention effect size seems to diminish at the 6-month FUP, there are still trends toward decreased anxiety and depression, and improvements in the HIP-10 score—a measure of self-efficacy and tendency toward making positive and intentional behavior choices.

The Pathways for Survivors workshop was based on an "intention model," which has been applied within numerous business and human resource settings and has been pragmatically refined over time. A pilot study among cardiac rehabilitation patients and their caregivers, also incorporating this "intention model," revealed more positive attitudes and an improved sense of control and hope related to health, which remained stable at FUP out to 12 weeks [24]. Multiple other positive psychology interventions including mindfulness, expressive writing, and creation of hope have been studied and found to have an overall favorable impact on the QOL of patients with breast cancer [25].

Although a formal mixed methods analysis to evaluate common themes of the feedback was not conducted, participants' comments reflected that they valued the toolkit of "setting intentions," exploring obstacles, and incorporating exercises in gratitude and recasting. Participants also rated the group experience as an important aspect of the workshop and reported that the opportunity to interact with other survivors, share experiences, and actively engage in discussions helped bring

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the concepts to life. Although this was a skills-based workshop, prior research has shown that breast cancer support groups and other forms of peer support provide emotional and informational benefits, although their short- and long-term impact on anxiety and depression is not fully proven [26,27].

Previous studies have supported the efficacy of CBT and mindfulness-based therapies in patients with cancer in addressing FCR, depression, anxiety, and QOL. A meta-analysis of cognitive behavioral interventions among patients with breast cancer undergoing active treatment reported that these techniques had a significant effect in reducing anxiety and depression, and reported that while therapy length or delivery did not significantly moderate the effect, individual therapy showed a slight trend toward eliciting better results on distress outcomes [13]. Another meta-analysis review of mindfulness-based stress reduction programs by Zhang et al [17] reported that most programs were 6-8 weeks long, and had significant effects on anxiety and depression. A randomized controlled trial including breast, prostate, and colorectal cancer survivors receiving 8 sessions of blended CBT revealed a significant decrease in FCR as well as anxiety and depression on the Hospital Anxiety and Depression scale at 3 months from baseline [16]. A pilot study of a 1-2–day psychosocial intervention combining mindfulness-based CBT and covering anxiety management and relationships or sexuality issues for young breast cancer survivors was well received and resulted in an overall gain in self-reported knowledge and confidence among participants [28]. This pilot study led to a much larger randomized controlled trial of a mindfulness-based program compared to survivorship education and a waitlist control for the management of depressive symptoms in younger breast cancer survivors. Our intervention is unique from many previously reported interventions in that it involves only a single session and is led by a lay moderator, making it more convenient and accessible to a population of patients who may find it challenging to attend multiple weekly sessions.

Limitations

Though the results support an improvement in anxiety, depression, and intention and resiliency as immediately as 1 week after the workshop, suggesting a direct impact of the intervention, as with many previously reported interventional studies that attempted to impact QOL in the survivorship population, this study was not a randomized trial. Nonetheless, we attempted to contextualize the Pathways participants, both in terms of clinical and demographic factors, and baseline anxiety and depression scores in comparison to a reference subpopulation of general early-stage breast cancer FUP patients. However, our routine care population was only sampled at one time point; therefore, we do not have a trajectory of their PROMIS anxiety and depression scores over time and thus does not serve as a true control group. Pathways participants were generally younger than both the average breast cancer survivor as well as our comparison group, and were of a high socioeconomic status (graduate degree holders, annual income of >US $100,000) and working on a full-time basis. As an academic center, we attract a higher risk and younger patient population. While we do see a diverse population, many of our patients are highly educated with high health literacy, who are seeking clinical trials, and are willing to participate in research. These patients are fit enough and have the financial resources to travel for their cancer care. Further research with more heterogeneous patients and with a larger 6-month follow-up sample is needed to confirm that the positive impact on several QOL measures from this positive psychology or mindfulness and skills-based workshop can be sustained and also observed in a more diverse population.

Clinical Implications

Transitioning from a patient with breast cancer to a breast cancer survivor is associated with a significant burden of psychological distress. Our study supports the Pathways for Survivors workshop as a highly satisfying and time-efficient means for breast cancer survivors to learn behavioral skills and the incorporation of this workshop into survivorship care may help improve emotional well-being and potentially overall QOL.

Conclusions

The Pathways for Survivors workshop was favorably received, and patients' anxiety and depression decreased significantly at 1 and 6 weeks after the workshop and remained below baseline at 6 months. While the 1-day workshop format is unique and is more convenient and accessible for patients who may find attending multiple weekly sessions challenging, future research is necessary to explore the impact of integrating of videoconferencing, additional "booster" sessions to reinforce the skills and concepts illustrated in this workshop, and to evaluate its longer-term impact.

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Authors' Contributions

M Melisko, GH, JH, DH, and ECW conceived and designed the study and analysis. SU, MEM, M Melisko, M Matthys, DH, EKH, and ECW collected the data. SU, M Matthys, JH, and M Melisko performed the analysis. SU, MEM, and M Melisko wrote the manuscript with the help of GH, ECW, AJC, HR, and LE.

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Conflicts of Interest

None declared.

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Abbreviations

BCC: Breast Cancer Center
CBT: cognitive behavioral therapy
FCR: fear of cancer recurrence
FUP: follow-up
HIP-10: Happiness Index Profile
PROMIS: Patient-Reported Outcomes Measurement Information System
QOL: quality of life
UCSF: University of California San Francisco

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Original Paper

Comparing Survivors of Cancer in Population-Based Samples With Those in Online Cancer Communities: Cross-sectional Questionnaire Study

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Abstract

Background: Most Western countries have websites that provide information on cancer and the opportunity to participate in online cancer communities (OCCs). The number of patients with cancer that participate in these OCCs is growing. These patients are relatively easy to approach for research purposes.

Objective: The objective of this study is to determine the differences and similarities between survivors of cancer in population-based samples and survivors participating in OCCs who use the internet in relation to their illness.

Methods: In 2017, we drew a sample of 539 population-based patients and 531 OCC patients. The population-based patients were sent a paper-based questionnaire, and the OCC patients were sent the same questionnaire on the web. In the questionnaire, we asked patients about their sociodemographics, internet use, sources of information, media use, and wishes regarding future internet use for health care–related purposes, and the effect of internet use on their health care consumption.

Results: The response rate of population-based internet users was 47% (233/496), and that of the OCC group was 40.3% (214/531). The OCC group had a significantly higher education level ($P<.001$), was younger ($P<.001$), had more survivors that were employed ($P<.001$), and attached greater importance to the internet (171/214, 79.9% vs 126/233, 54.1%; $P<.001$) and fellow survivors (107/214, 50% vs 60/233, 25.8%; $P<.001$). Compared with the population-based group, the OCC group reported more intensive internet use immediately after diagnosis, during treatment, and during follow-up ($P<.001$ in each case). There were similarities in terms of the relative importance that survivors attach to the various sources of information, the topics on which they seek information, and their wishes for future eHealth possibilities. The OCC group reported a greater need to participate in a web-based class or chat with others (92/214, 43% vs 44/233, 18.9%).

Conclusions: We conclude that survivors who are members of an OCC are not representative of survivors of cancer in general. There are significant differences in sociodemographic characteristics, internet use during their treatment journey, internet search frequency during their cancer journey, and participation wishes. Using web-based information and communication can support shared decision-making and may facilitate the active participation of patients during their treatment. For research purposes, it is important to take the bias in OCC groups into account.

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KEYWORDS

internet use; breast cancer; prostate cancer; lymphoma; gynecological cancer; cancer survivors; online health community

Introduction

Background

Over the past decade, an increasing number of people have been using the internet, especially in Western countries such as the Netherlands, where the availability of the internet is very high [1,2]. Many countries have websites that provide not only information on cancer but also the opportunity to participate in an online cancer community (OCC) or be a member of a web-based cancer platform. For example, in the Netherlands, there is Kanker.nl [3]; in the United Kingdom, there is Macmillan [4]; and, in the United States, there is the American Cancer Society (the related community [5]) [6]. On these websites, patients can find information about the various types of cancer and their treatment or treatments, side effects, and long-term effects. Visitors can also create a profile and become members to read, start a blog, or communicate with fellow patients through chat groups and personal messages. Members of such communities or platforms are often asked to be respondents in cancer research [7-9]; however, to the best of our knowledge, there have been no studies that have systematically compared survivors of cancer with a profile in OCCs with those in population-based samples. Are the characteristics, internet use, and wishes of Dutch survivors of cancer who participate in an OCC different from a selection of survivors of cancer from the Netherlands Cancer Registry (NCR)?

Previous studies on internet use among patients with cancer have shown that the number of patients who use the internet for information, communication, and community purposes has increased sharply in recent years [10-12]. However, the topics that interest patients have remained more or less stable over the same period [10]. Differences between patients over time have been found in the extent to which they use the internet. These have been attributed to (1) gender (men use the internet more often than women), (2) age (young people use the internet more than older adults), and (3) education level (highly educated people use the internet more than those with a low level of education) [10,13-17]. Research has shown that women tend to participate in OCCs more often than men [15,18]. The explanation often given is that women are more often caregivers [19], are more active in health issues, and have different needs for emotional support than men [20-22].

Despite patients' increasingly intensive use of the internet, health care professionals are still their most important source of information [10,23]. In recent years, much has changed in the physician–patient relationship [24]. The former, predominantly paternalistic approach has made way for a more patient-centered approach with attention to shared decision-making and patients' individual wishes [24]. When a patient with cancer is confronted with late effects and is chronically affected by it, the *patient-as-partner* concept may be most appropriate, whereby the patient is a participating member of the treatment team [24]. To become a partner, a patient must first develop learning, then assessment, and ultimately adaptation practices [25].

The internet may actively contribute to shared decision-making and patient-as-partner practices as patients can use it independently from their health care professionals; it is always available; and it offers every individual option for content, communication, and community involvement. Researchers frequently recruit and look at patients with cancer who participate in OCCs to find out to what extent patients with cancer have these skills. However, to what extent are these patients representative of the entire population of patients with cancer?

Objective

In this study, we aim to identify the differences and similarities between survivors of cancer who participate in an OCC and population-based samples of survivors of cancer who use the internet in relation to their illness. We believe it is important to know the differences between these 2 groups as many studies are based on data from survivors in the OCC group, which raises the important question of the extent to which these findings generalize to the complete population of survivors of cancer [7-9]. Although this is an important methodological question, it has received very little attention. We hypothesize that there are significant differences between these 2 groups. First, we expect that survivors in the OCC group who use the internet have different sociodemographic characteristics compared with survivors in the population-based group. Second, we expect that survivors in the OCC group use the internet more often and have different wishes for various purposes, including content, communication, community, and eHealth, compared with survivors in the population-based group. Finally, we expect that survivors in the OCC group are more active media users for communication with health care professionals and relatives than survivors in the population-based group.

Many definitions of cancer survivorship have been used. In this paper, we chose to adopt the most frequently used definition that is also applied by the US National Coalition for Cancer Survivorship and Institute of Medicine: "a person is considered to be a cancer survivor from the time of diagnosis through the balance of his or her life" [26].

Methods

Ethics Approval

A declaration of no objection was granted by the medical ethics review committee Midden Brabant (NW2016-47).

Participants

For the population-based group, a population-based, cross-sectional survey on internet use was conducted through the NCR. In October 2016, we drew a random sample of 523 patients with breast cancer (138/523, 26.4%), prostate cancer (125/523, 23.9%), gynecological cancer (184/523, 35.2%), or lymphoma (76/523, 14.5%) diagnosed in 4 hospitals in the

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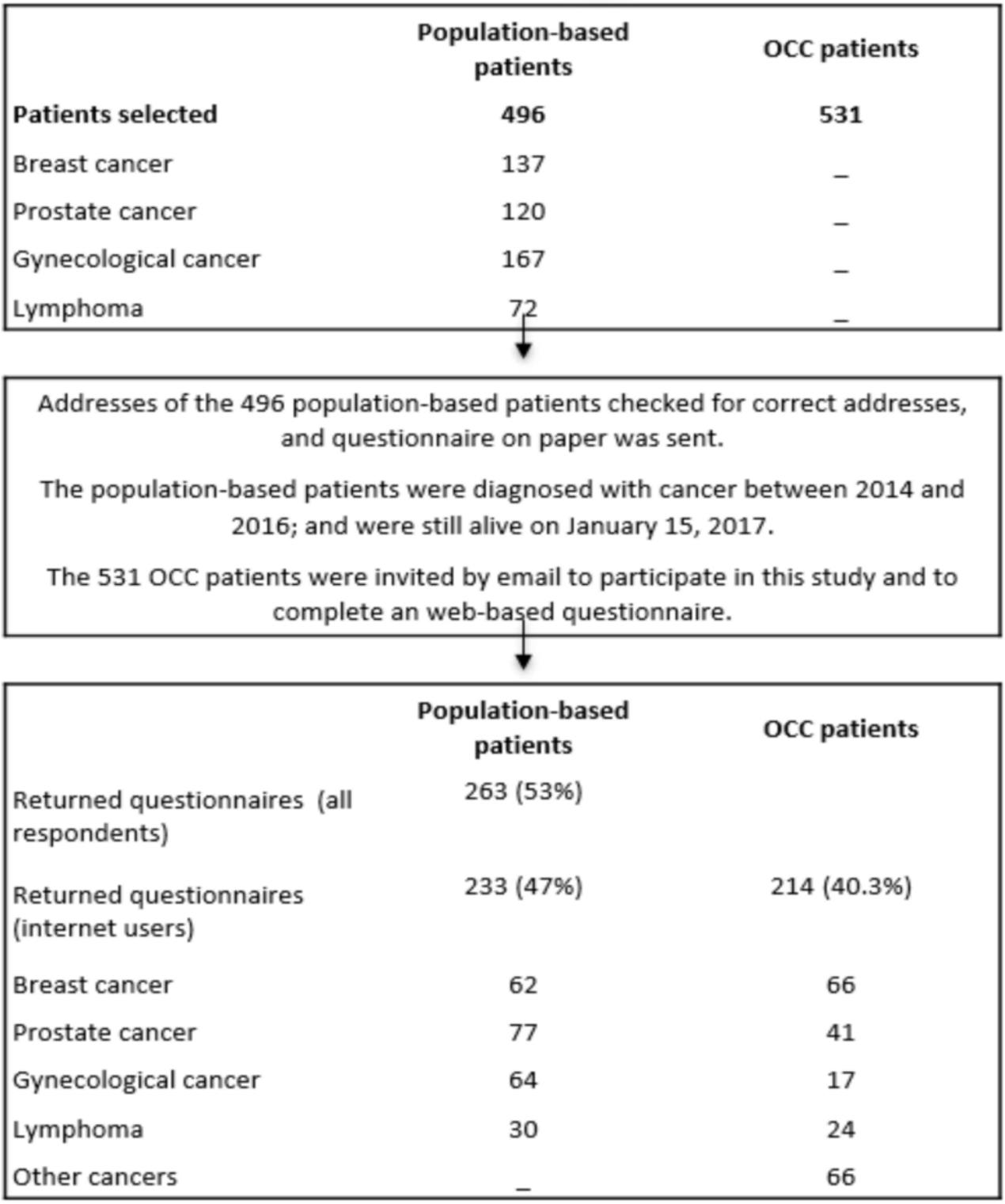
period between 2014 and 2016 and who were aged ≤70 years at diagnosis. Our samples were linked with the Dutch municipal records database that contains mortality and residential data from all citizens through municipal registries to exclude all deceased patients. Addresses were checked for correctness, and all 496 surviving patients were sent an information letter together with a paper and pencil questionnaire by their oncologist. By replying, the patients explicitly agreed to participate and consented to the linkage of their questionnaire data with their disease history as registered in the NCR. The returned questionnaires were only identifiable by a study number, which guaranteed patient anonymity. We repeated the research method from 2005 to 2017 to describe the changes over time [10]. For the full selection procedure, see Figure 1 and the flowchart in the paper by van Eenbergen et al [10]. For this study, we included only the population-based participants who used the internet.

Figure 1. Flowchart of the data collection process. OCC: online cancer community.



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For the OCC group, in 2017, we approached members of the Kanker platform who indicated that they wanted to participate in research. We selected members with one of the following types of cancer: breast cancer, prostate cancer, gynecological cancer, lymphoma, colon cancer, rectal cancer, lung cancer, melanoma, or esophagus cancer (n=531). Kanker is the only web-based platform in the Netherlands for survivors of cancer and their relatives, where they can find trusted medical content and user-generated content. Kanker is an initiative of the Dutch Cancer Society (KWF Kankerbestrijding), the Dutch Federation of Cancer Patient Organisations, and the Netherlands Comprehensive Cancer Organisation. The platform started in 2013 with the functions of content, communication, and community. In 2020, Kanker had >500,000 visitors per month and approximately 32,500 members (July 2020). Members can make contact to communicate with other survivors and relatives, start a blog (1100 bloggers), participate in web-based discussion groups (50 groups), or participate in the research panel (1500 members). Visitors have to become members of Kanker for reading or posting user-generated content. The medical information is checked by professionals. To help the users generate content, Kanker has peer moderators.

The population-based group patients were asked by their physician to participate in the study and complete a questionnaire on paper. OCC members who indicated in their membership profile whether they were willing to complete questionnaires and who met our selection criteria (survivor and cancer type) were invited by the community manager of Kanker by email to participate in this study. This email contained a link to a web-based questionnaire.

As their names and addresses were unknown, a paper questionnaire could not be sent to the OCC group. The population-based group filled in an informed consent form before completing the questionnaire. Through an opt-in option in the Kanker terms of use, the OCC group gave their (informed) consent so that they could be approached to request their participation in the study.

Measures

The NCR routinely collects data on tumor characteristics such as date of diagnosis; subsite; histology; stage (TNM clinical classification); primary treatment; and patient characteristics, including sex and date of birth. Kanker.nl respondents were asked to indicate certain tumor characteristics in the questionnaire (Multimedia Appendix 1 [15]; questionnaire translated; questions A, B, and C).

As no validated Dutch questionnaire on internet use among patients with cancer existed, we developed one in 2004, which was reviewed by an expert panel of 3 researchers and 6 survivors of cancer [27]. This questionnaire was based on the four areas of internet use—content, communication, community, and eHealth—defined by Eysenbach [28]. In 2017, we updated some of the questions because of internet developments in the intervening years, including increased access to Kanker, eHealth, social media, and blended care [10] (Multimedia Appendix 1; questionnaire translated; questions 27 and 29-42).

We used the same questionnaire for both groups; the population-based group filled out this questionnaire offline, and the OCC group did so on the web. The number of survivors in the population-based group on the web was unknown, and all the OCC group members were active on the web. In the questionnaire, we asked patients about their sociodemographics, internet use, sources of information for health care–related purposes, wishes regarding future internet use for health care–related purposes, self-management skills, and the effect of internet use on their health care consumption.

Statistical Analysis

All statistical analyses were performed using the SPSS statistical software (version 24.0; IBM Corp). Data regarding patient characteristics were compared between the population-based and OCC groups using chi-square analyses for categorical variables and independent-sample, 2-tailed *t* tests for continuous variables (Table 1). Chi-square analyses were conducted to investigate differences between the population-based and OCC groups in (1) information sources found to be important (Table 2), (2) distributions of search frequencies for each different disease phase (Figure 2), and (3) effects of internet use and participation in OCCs (Multimedia Appendix 2, Table S1). Finally, separate multivariate logistic regression analyses were conducted to investigate the independent association between the type of population (population-based group vs OCC group) and internet search frequency (outcome) treated as a dichotomous variable (daily or several times a week vs several times a month or year, or never) while adjusting for patient (age, gender, and education) and disease (time since diagnosis) characteristics (Multimedia Appendix 2, Table S2). The tests were 2-sided, considered statistically significant at $P<.05$, and adjusted for multiple testing using the Bonferroni correction.

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Table 1. Patient characteristics separated by type of patient group.

Characteristic	Netherlands Cancer Registry (population-based)	Kanker (OCC[a])	*P* value
Patients selected, N	523 (100)	531 (100)	N/A[b]
Returned questionnaires[c], n (%)	233 (44.6)	214 (40.3)	N/A
Gender, n (%)			.66
Female	142 (60.9)	126 (58.9)	
Male	91 (39.1)	88 (41.1)	
Age at time of survey (years), n (%)			<.001
<50	39 (16.7)	41 (19.2)	
50-65	98 (42.1)	130 (60.7)	
>65	96 (41.2)	43 (20.1)	
Age at time of survey (years), mean (SD)	61.8 (11.6)	58.1 (9.5)	<.001
Tumor, n (%)			<.001
Breast cancer	62 (26.6)	66 (30.8)	
Prostate cancer	77 (33)	41 (19.2)	
Gynecological cancer	64 (27.5)	17 (7.9)	
Lymphoma	30 (12.9)	24 (11.2)	
Other cancers[d]	N/A	66 (30.8)	
Months since diagnosis, n (%)			<.001
0-18	6 (2.6)	51 (23.8)	
19-24	50 (21.5)	12 (5.6)	
25-30	74 (31.8)	15 (7)	
31-42	103 (44.2)	135 (63.1)	
Months since diagnosis, median	29	42	<.001
Months since diagnosis, mean (SD)	30.2 (6.9)	55.5 (59.0)	<.001
Education[e], n (%)			<.001
Primary school	43 (18.6)	11 (5.1)	
Secondary school	114 (49.4)	94 (43.9)	
College or university	75 (32.5)	108 (50.5)	
Employment status[f], n (%)			<.001
Employed (ill)	94 (40.9)	124 (57.9)	
Employed (on insurance)	25 (10.8)	70 (33.2)	
Unemployed	136 (59.1)	90 (42.1)	
Marital status[g], n (%)			.03
Married or living together	191 (82)	174 (82.1)	
Partner, not living together	13 (5.6)	3 (1.4)	
No partner	28 (12)	36 (17)	
Children[h], n (%)			.003
None	33 (14.2)	50 (23.4)	
Yes, living with one or both parents	40 (17.2)	51 (23.8)	
Yes, living somewhere else	159 (68.5)	113 (52.8)	

[a]OCC: online cancer community.

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[b]N/A: not applicable.
[c]Only internet users.
[d]Including colon cancer, rectal cancer, lung cancer, melanoma, esophagus cancer, and other.
[e]Missing for 2 patients.
[f]Missing for 3 patients.
[g]Missing for 2 patients.
[h]Missing for 1 patient.

Table 2. Sources of information found to be important (N=447).

Source of information	Ranking		Population-based (n=233), n (%)	OCC (n=214), n (%)	*P* value
	Population-based	OCC[a]			
Medical oncologist	1	1	212 (91)	189 (88.3)	.35
Oncology nurse	2	3	154 (66.1)	154 (72)	.71
Internet for information	3	2	126 (54.1)	171 (79.9)	<.001
Family	4	6	120 (51.5)	84 (39.2)	.009
Friends	5	7	115 (49.4)	76 (35.5)	.003
General practitioner	6	5	100 (42.9)	94 (43.9)	.83
Children	7	11	97 (41.6)	65 (30.4)	.01[b]
Other patients	8	4	60 (25.8)	107 (50)	<.001[b]
Other patients via the internet	15	10	17 (7.3)	68 (31.7)	<.001[b]
Group discussions with patients	17	8	13 (5.6)	70 (32.7)	<.001[b]
Colleagues	11	15	41 (17.6)	32 (15)	.45[b]
Pharmacist	9	14	48 (20.6)	40 (18.7)	.61[b]
Newspapers or television	9	12	49 (21)	63 (29.4)	.04
Books	11	8	41 (17.6)	71 (33.2)	<.001
Second-opinion physician	13	13	20 (8.6)	56 (26.2)	<.001
Alternative counselor	14	17	18 (7.7)	30 (14)	.03
Home care nurse	15	15	16 (6.9)	32 (15)	.006

[a]OCC: online cancer community.
[b]A relatively large difference in ranking (≥4).

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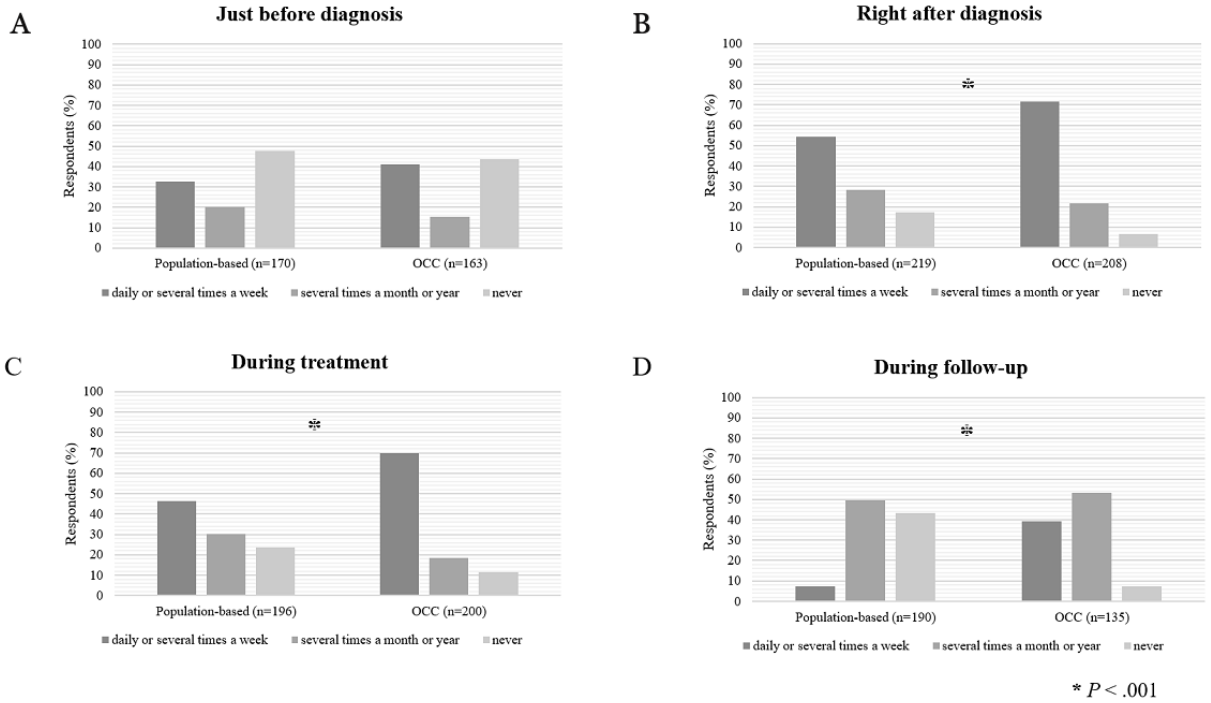
Figure 2. Internet search frequency for information on cancer just before diagnosis (A), right after diagnosis (B), during treatment (C), and during follow-up (D). OCC: online cancer community.



Results

Overview

The two groups showed similar response rates: 47% (233/496) for the population-based group and 40.3% (214/531) for the OCC group. In the OCC group, 30.8% (66/214) had a cancer type other than lymphoma, prostate cancer, breast cancer, or gynecological cancer. As we found no significant differences between the results for patients with different cancer types, we only report the totals.

Patient Characteristics

Differences between the 2 groups were evident with regard to patient characteristics (Table 1). Compared with the population-based group respondents, the OCC group respondents had a higher education level (college or university: 108/214, 50.5% vs 75/231, 32.5%; *P*<.001) and were younger (mean age 58.1, SD 9.5 years vs 61.8, SD 11.6 years; *P*<.001), and more respondents were employed (124/214, 57.9% vs 94/233, 40.3%; *P*<.001). Compared with the population-based group, the OCC group respondents had children less often (164/214, 76.6% vs 199/232, 85.7%; *P*=.003).

Internet Use

The following results for questions about participation in an OCC were reported by the OCC and population-based group respondents, respectively: reading posts of other survivors (120/214, 56.1% vs 26/114, 22.8%; *P*<.001), creating a profile (158/214, 73.8% vs 16/114, 14%; *P*<.001), and actively posting text in a blog or a discussion group (35/214, 16.4% vs 6/114, 5.3%; *P*<.001; Multimedia Appendix 2, Table S3). Overall, the population-based group hardly participated in a web-based health community.

Regarding communication and social media known in 2017, the OCC and population-based group respondents mainly used email and WhatsApp to communicate about their illness with family members (148/214, 69.2% vs 149/233, 63.9%), children (101/214, 47.2% vs 123/233, 52.8%), friends (158/214, 73.8% vs 142/233, 60.9%), and their oncologist (73/214, 34.1% vs 61/233, 26.2%). Facebook and blog posts were used more often to communicate with fellow survivors (110/214, 51.4% vs 28/233, 12%). The other available media—Twitter and Skype—were rarely or never used. The OCC group reported more intensive use of digital media and maintained web-based contact with a greater variety of people (Multimedia Appendix 2, Table S4).

The OCC group respondents were less satisfied with the information they had received than the population-based group (131/214, 61.2% vs 200/233, 85.8%; *P*<.001). The OCC group attached greater importance to all information sources except family members and children (if any) than the population-based group. Most of the differences in the importance of the information sources were statistically significant, including the internet (171/214, 79.9% vs 126/233, 54.1%; *P*<.001), fellow survivors (107/214, 50% vs 60/233, 25.8%; *P*<.001), and mass media (63/214, 29.4% vs 49/233, 21%; *P*=.04; Table 2).

In their ranking of information sources on relative importance, there were many similarities between the population-based and OCC groups except for the importance that patients attached to fellow patients and their own children.

In almost all phases of the patient journey during the illness, the OCC group reported more intensive internet use (Figure 2). The differences in the three phases were significant: (1) immediately after diagnosis, (2) during treatment, and (3) during follow-up (*P*<.001 in each case). Only just before diagnosis, the distribution of internet use between the 2 groups did not

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differ significantly. These results were also found when adjusting for patient (age, gender, and education) and disease (time since diagnosis) characteristics (Multimedia Appendix 2, Table S2). In addition, the population-based group indicated *not applicable* more often in the *during treatment* phase, which suggests that the population-based group respondents received treatment less often, the difference being 16% (109/214, 50.9% vs 82/233, 35.2%). The population-based respondents reported being in the *follow-up* phase more often (189/233, 81.1% vs 135/214, 63.1%), which did not result in more intensive internet use in that phase. The population-based respondents were probably less seriously ill; thus, fewer treatments were needed to enter the follow-up phase. The OCC group underwent more treatments, and most are still in the treatment phase (Multimedia Appendix 2, Table S5).

In searching for information on all topics included in the questionnaire, the OCC group reported using the internet more intensively than the population-based group, the mean difference being 23%. Searching for information on *cancer support groups*, *trials/research*, and *type of cancer* diverged strongly from that mean (by 42%, 37%, and 13%, respectively). To determine whether both groups found the same topics important, the percentage for each group was used to rank the topics from 1 to 18. The 2 groups ranked nearly all topics equally on importance, except for *consequences for sexuality* (7 vs 12, respectively) and *cancer support groups* (13 vs 9, respectively; Multimedia Appendix 2, Table S6).

More survivors in the OCC group reported that after using the internet, they were better informed (92/214, 43% vs 68/233, 29.2%; P=.002) to discuss the information with their physician more often than the population-based group (21/214, 9.8% vs 9/233, 3.9%; P=.004). There were no differences in terms of whether the information they had obtained influenced their choice of treatment (45/214, 21% vs 37/233, 15.9%; P=.14). Neither group reported that their internet use led to more consultations with a physician (2/214, 0.9% and 2/233, 0.9%; Multimedia Appendix 2, Table S1).

Wishes Regarding Internet Use

For all topics, survivors' wishes with regard to internet use exceeded current possibilities (Table 3). The 2 groups reported similar use of resources on all topics. Their use at the time of completing the questionnaire differed by a mean of 5%, whereas the wishes regarding all topics differed by a mean of 16%.

For both groups, the difference between possibilities and wishes was greatest for *getting advice on supportive health care* (possibilities: 0%; wishes: 126/233, 54.1% and 148/214, 69.2%). The OCC group reported 24% higher wishes related to participating in a web-based self-management class and chatting with others (44/233, 18.9% vs 92/214, 43%).

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Table 3. Patients' current use of and future wishes for internet possibilities (N=447).

Item	Current use		Future wishes		Ranking wishes	
	Population-based (n=233), n (%)	OCC[a] (n=214), n (%)	Population-based (n=233), n (%)	OCC (n=214), n (%)	Population-based	OCC
Accessing own test results	72 (30.9)	77 (36)	170 (73)	184 (86)	1	1
Accessing own medical file	75 (32.2)	77 (36)	165 (70.8)	182 (85)	2	2
Making an appointment	56 (24)	81 (37.9)	161 (69.1)	173 (80.8)	3	3
Requesting prescriptions	72 (30.9)	81 (37.9)	156 (67)	165 (77.1)	4	4
Getting personal advice on symptoms	N/A[b]	N/A	142 (60.9)	152 (71)	5	5
Emailing with oncologist	58 (24.9)	64 (29.9)	135 (57.9)	152 (71)	6	5
Getting advice on supportive care	N/A	N/A	126 (54.1)	148 (69.2)	7	7
Receiving reminders	56 (24)	56 (26.2)	123 (52.8)	143 (66.9)	8	9
Making complaints	58 (24.9)	88 (41.1)	123 (52.8)	139 (65)	8	11
Emailing with nurse	82 (35.2)	73 (34.1)	119 (51.1)	146 (68.2)	10	8
Self-monitoring of treatment consequences	N/A	N/A	112 (48.1)	143 (66.8)	11	9
Rating health care professionals or hospitals	N/A	N/A	93 (39.9)	116 (54.2)	12	14
Requesting tests	28 (12)	17 (7.9)	105 (45.1)	118 (55.1)	13	13
Suggesting ideas	35 (15)	47 (22)	96 (41.2)	139 (65)	14	11
Requesting referrals	21 (9)	28 (13)	96 (41.2)	111 (51.9)	14	15
Performing self-diagnosis tests	7 (3)	9 (4.2)	63 (27)	88 (41.1)	16	18
Participating in web-based self-management class	N/A	N/A	44 (18.9)	92 (43)	17	16
Requesting oncologist via forum	7 (3)	6 (2.8)	44 (18.9)	81 (37.9)	17	19
Chatting with others	9 (4)	11 (5.1)	42 (18)	92 (43)	19	16
Asking questions of an oncologist in forum	5 (2)	9 (4.2)	35 (15)	73 (34.1)	20	20

[a]OCC: online cancer community.

[b]N/A: not applicable.

Discussion

Principal Findings

Dutch survivors participating in a web-based cancer community (the OCC group) were younger, more educated, more likely to be employed, and more likely to be unemployed because of illness than the population-based group. Significantly fewer members of the OCC group had children, and they found fellow survivors and web-based group discussions relatively more important as sources of information than their close relatives.

Differences in Patient Characteristics

Approximately 69.1% (148/214) of the OCC group were survivors of the same 4 cancer types as the survivors in the population-based group. The remaining 30.8% (66/214) were survivors of 6 other random cancer types. Our additional analyses demonstrated that information needs and internet use were not influenced by cancer type. This can be confirmed by previous studies that showed that information seeking and illness-coping styles seem to influence how patients process information [29,30].

To increase the reach among the average population, we decided to repeat our research method of 2005 and asked the population-based group to complete the questionnaires on paper. Importantly, earlier research has shown that there is no difference in response rate between different invitation modes [31,32]. In this study, we show that there are differences between the population-based and OCC groups, not only in terms of sociodemographic characteristics. The OCC group seemed to have undergone more treatments (Multimedia Appendix 2, Table S5). The OCC group may experience more late effects of their treatment and seem to have less control over the consequences of their disease and treatment. The active involvement in Kanker.nl suggests that they hope that change is still possible.

Differences in Internet and Media Use

This study revealed significant differences in internet use between the population-based and OCC groups. The latter searched for information on clinical trials markedly more often. A possible explanation for this phenomenon, as indicated by previous studies, is that younger and highly educated respondents tend to search for such information more often and

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tend to understand it better than older respondents with a low level of education [33,34].

As far as we have been able to ascertain, only a limited portion (<25%) of the population-based group respondents participated in an OCC [10]. The OCC group found fellow survivors significantly more important as a source of information, which is probably why they participate in an OCC. Fellow survivors provide both emotional and informational support [15].

The OCC group respondents communicated more often with oncologists (73/214, 34.1% vs 61/233, 26.2%) and fellow survivors (110/214, 51.4% vs 28/233, 12%) than their population-based group counterparts and used more different media to interact with their social network in relation to their illness (Multimedia Appendix 2, Table S4).

These differences require not only access to information but also possession of health-related skills such as the ability to formulate meaningful questions [24,35,36]. Actively using the internet to access information, participate in an OCC, and communicate with their social network enables survivors to develop those skills [37]. Recent studies have shown that participating in such a community makes survivors more resilient, which also enables self-management [38,39].

Differences in Wishes Regarding Internet Use

The 2 groups reported different wishes, although the ranking of the wishes in order of importance was markedly similar. This is in line with our previous study comparing internet use of survivors in the population-based group in 2005 and 2017 [10], which showed that the intensity of use changed with time, although the ranking of wishes remained stable.

Many of the survivors' wishes were related to eHealth, which makes it possible for them to actively participate in illness and recovery management. An important aspect is access to their own electronic health record (EHR). According to the Netherlands' eHealth monitor 2018, approximately 45% of citizens had access to their EHR [40]. This corresponds roughly to the use of their own medical file reported by OCC respondents. EHR use by the population-based respondents ranged from 24.9% (58/233) to 35.2% (82/233) in this study.

A possible explanation could be that the intensive internet users—in this study, the OCC group—are probably early adopters of eHealth. They would seem to be accurate indicators of future internet use by a large number of survivors [10]. If so, then in the coming years, eHealth interventions will be increasingly used to self-monitor one's own illness management behavior. This effect may be amplified as more patients with cancer survive longer, often with more long-term and late effects.

Differences in Treatment and Sense of Control

The OCC group underwent more treatments. It seems understandable that these survivors experience the consequences of treatment more and have an insufficient sense of control over these symptoms. The survivors actively searched for information and joined an OCC (Figure 2). The population-based group had fewer treatments and, therefore, fewer problems coping with their symptoms compared with the OCC group [41,42]. The OCC group had more reasons to investigate what could possibly help them, in which case eHealth tools for self-care are an accessible option [43].

The OCC group has the characteristics of patients with chronic disease [44,45]. For them, *patient-as-partner* is the most appropriate concept [24,25]. The more active attitude is confirmed in their more frequent internet searches on topics such as *trials/research*, *cancer support groups*, and *What can I do myself?* Within the possibilities, they also make greater use of eHealth and have more wishes for future active participation in their health situation, such as shared decision-making, monitoring side effects, and seeking personal advice. It is unclear whether the OCC group comprises survivors who less readily accept the consequences of their illness or are more aware of them or are less able to cope with them or expect their symptoms to diminish. They probably expect that they can improve their health through active participation and self-management. Indeed, the characteristics of the survivors in the OCC group are factors that influence the self-management of individuals in an eHealth environment [43]. Could this OCC group represent the starting point for user uptake and implementation of web-based interventions, many of which remain on the shelf [46,47]? It may be that eHealth feels too burdensome for survivors and that the interventions should be more focused on e-Learning. An example of this is the cancer support community in the United States [48], which is less stigmatizing and appeals to people's motivation in combination with their abilities to learn and communicate. Follow-up research into the web-based wishes of OCC participants could be directed at determining to what extent this growing group of patients with chronic cancer is motivated to take a course through a web-based patient academy that appeals to people's skills and possibilities.

Limitations of the Study

This study has several limitations that need to be addressed. First, we approached and surveyed the 2 sample groups in different ways. The population-based group respondents were asked by a physician to participate in the study and completed a paper-based questionnaire, whereas the OCC group was invited to participate on the web through the Kanker platform. For the latter group, we knew neither who their physicians were nor where they lived. We could not send them a paper questionnaire, so they answered the questions on the web. Studies on the use of web-based questionnaires versus paper questionnaires show that these methods can be used side by side [31,49]. Although these different research methods are unlikely to cause differences in results, we are not sure whether our sample is fully representative of OCCs. It may be that the members included in this study were the more active users of the OCC. However, this active group will likely correspond to the group of survivors that researchers have access to.

Furthermore, the population-based group included a small group of respondents who actively participated in an OCC such as Kanker. We did not consider this as an exclusion criterion as in any population-based sample, there are survivors who participate in a web-based community. The differences between

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the 2 groups would have been larger if we had excluded these respondents.

A final limitation is that the study was conducted only in the Netherlands, where internet access is extremely high, and the respondents have an above-average education level. Although the typical Dutch survivor of cancer may be different in certain ways from those in other Western countries, previous studies have shown that there are many similarities between the web-based behavior of survivors in various countries [14-16,18].

Conclusions

We conclude that survivors who participate in an OCC (both posters and lurkers) are not representative of survivors of cancer in general. There are significant differences in (1) sociodemographic characteristics, (2) internet use during their treatment journey, (3) internet search frequency during their cancer journey, and (4) participation wishes. However, there are also certain similarities in terms of the relative importance that survivors attach to the various information sources, the topics on which they seek information, and their wishes for future eHealth possibilities. Any differences in importance ranking can be attributed to the OCC group being an internet-based community that actively seeks contact with fellow survivors.

The above findings and conclusions have implications for other researchers. Most importantly, if they recruit study participants through an OCC, they will not be fully representative of the general patient population. Arguably, an OCC group is more suitable for research into supportive care in relation to survivorship. The survivors in the OCC group experience long-term effects and seem motivated to gain a sense of control over them, which could be a good motivational factor to participate in web-based intervention studies. In general, it is advisable to take the specific nature of an OCC sample into consideration when reporting findings for this particular group of survivors of cancer.

In general, we recommend that survivors of cancer use internet resources throughout their illness and treatment journey. There are differences between the 2 groups because of the circumstances in which they find themselves; however, the internet offers different options for different circumstances. The wishes are similar; however, the use differs, which could be explained by age, gender, number of treatments, and communication needs.

Web-based information and communication can support shared decision-making and may facilitate the active participation of patients during their treatment. At the start of that journey, they have a great need for information, which is essential for shared decision-making [36]. After cancer treatment, such a platform provides patients with chronic cancer with an environment that seems to facilitate their active participation in their treatment [24,39].

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Authors' Contributions

All authors have read and approved the contents of this manuscript.

Conflicts of Interest

None declared.

Multimedia Appendix 1
Questionnaire translated.
[DOCX File , 117 KB - cancer_v8i1e19379_app1.docx]

Multimedia Appendix 2
Tables with additional data.
[DOCX File , 33 KB - cancer_v8i1e19379_app2.docx]

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Abbreviations

EHR: electronic health record
NCR: Netherlands Cancer Registry
OCC: online cancer community

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Original Paper

An Analysis of Health Care Team Communication Needs Among Younger vs Older Breast Cancer Survivors: Web-Based Survey

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Abstract

Background: Prior studies indicate that the age of onset of breast cancer is an important element in considering communication between patients and the health care team. Younger women aged 45 and under diagnosed with breast cancer are often at a higher risk of being more vulnerable to psychosocial issues compared to older women aged 46 years and above. Few studies have examined age differences in patient perceptions of treatment-related discussion and communication during transition with their health care team.

Objective: The aims of this survey were (1) to better understand breast cancer survivors' perspectives regarding communication with health care providers during treatment and during transition to posttreatment care; and (2) to determine the differences between younger women with breast cancer (≤45 years of age) and older women (≥46 years of age). It was hypothesized that (1) breast cancer survivors' psychosocial and finance-related communications with health care providers may lack effectiveness; (2) younger women experience greater needs for patient-centered communication with physicians and health care providers, especially about psychosocial care and transition to posttreatment care; and (3) younger breast cancer patients (≤45 years of age) need more information on survivorship and follow-up care.

Methods: An internet-based survey was conducted with 143 women in Central Texas with 35% (n=50) aged 45 years or under and 65% (n=93) aged 46 years and above. The Mann-Whitney *U* test was performed to assess differences in participants' perceptions about communication with health care providers by age group: younger (≤45 years of age) and older (≥46 years of age) women.

Results: Statistically significant results pertained to rating health care team and patient discussions about transition from treatment to posttreatment using scores of 0 as "no discussion" and 100 as "in-depth discussion." For the questions about management of posttreatment care, the overall mean score of the groups was 56.26 and that of the younger group was 43.96; the mean score of the older group was 61.96 (P=.02). For the question about the timing of follow-up appointments, the overall mean score was 64.29; the mean score of the younger group was 54.44, and that of the older group was 68.88 (P=.05). All the group scores related to psychosocial and financial support discussions with health care providers were low, with a rollup average of only 30.02 out of 100, suggesting that this is an important area for improving patient-centered communication.

Conclusions: For all patients, transition from treatment to posttreatment requires a greater level of engagement and communication with the health care team. It appears that younger patients aged ≤45 years require more in-depth and personalized messaging to better understand their posttreatment care requirements.

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KEYWORDS

breast cancer; breast cancer survivorship; patient-physician communications; patient-centric communication; younger breast cancer patients; patient communication

Introduction

Although the median age at presentation is approximately 62 years in the United States [1], approximately 11% of all breast cancers occur in women younger than 45 years [2]. Breast cancer survivors diagnosed at younger ages are confronted with multiple demands of managing families and careers as well as complex medical, psychosocial, and behavioral late effects, including fertility and mental health issues. They may also be dealing with financial toxicity as a result of their treatment and may lack health benefits including sick leave and paid time off [3,4].

Compared to older women, younger women generally have more aggressive cancers, lower survival rates, and are more likely to experience recurrence of cancer [5-10]. Communication between patients and their health care teams is critical for the delivery of high-quality, patient-centered care, and it is associated with improved adherence to posttreatment protocols, patient satisfaction, and self-management [11,12]. Health care team members may include oncologists, nurses, social workers, and patient navigators.

In this study, we used an internet-based survey tool to better understand the differences in the perceptions on health care provider team support between younger and older breast cancer survivors during treatment and the transition from treatment to posttreatment care. We hypothesized that (1) there may be breakdowns in communication during treatment and transitions in care between breast cancer and health care providers and (2) that younger women (ie, aged ≤45 years) have greater needs for patient-centered communication, especially that related to psychosocial care and the transition to posttreatment care. An additional hypothesis was that younger breast cancer patients (<45 years of age) need more information on survivorship and follow-up care during transition, as they are expected to live longer and may also have a higher rate of recurrence [12].

Methods

Overview

The Breast Cancer Resource Center (BCRC) was the recipient of a 5-year cooperative agreement with the US Center for Disease Control (CDC) under a grant entitled “Multiple Approaches to Support Young Breast Cancer Survivors and Metastatic Breast Cancer Patients.”

The grant is focused on improving services and access to resources for young breast cancer survivors diagnosed under the age of 45 years and for metastatic breast cancer patients. A multifaceted needs assessment, consisting of focus groups, key informant interviews, and an internet-based survey, was conducted to determine what unmet needs exist in Central Texas.

Participants were able to access it via the internet from a link provided by BCRC to its constituents and collaborating organizations from August to November 2020. There were no incentives for participants who completed the survey. Results were screened for automated agents or bots and duplicate entries. BCRC sought institutional review board approval from the University of Texas at the Austin Office of Research Support and Compliance and was granted an exemption (reference: FWA # 00002030).

Recruitment

The participants were recruited using convenience sampling via email from a number of Texas-based cancer and breast cancer advocacy groups. The Cancer Alliance of Texas was also involved in the recruitment process by distributing surveys among its participating organizations, agencies, institutions, and individual members. BCRC formed an advisory council, consisting of physicians, researchers, stakeholders, and survivors, to support the CDC grant activities, including assisting in dissemination of the survey. The survey was also advertised on the BCRC Facebook pages, and the link was emailed to anyone who had been a BCRC client since 2018. The web-based survey was a voluntary, open survey, and it was created and distributed by BCRC to breast cancer survivors in Texas who had a previous or current breast or metastatic breast cancer diagnosis.

Survey Design

The survey consisted of 44 questions and was created using Qualtrics (Qualtrics International). Content from earlier focus groups and key informant interviews informed the questions included in the survey and the draft survey was tested with survivors and members of the project’s advisory group. Anonymous responses were captured directly in Qualtrics, and they were later downloaded directly from the software for analysis.

Survey participants were informed of the average length of time the survey would take, the purpose of the survey, how the responses would be used, and who the investigator was. Adaptive questioning was used to reduce the number and complexity of the questions. Survey participants could go back and review their responses before submitting. The survey collected various demographic data including ethnicity, education level, and income and insurance status. Women were asked to rate the helpfulness of their health care team, as it related to aspects of treatment. Participants were queried about their concerns regarding treatment-related issues and the level of discussion with their health care providers about treatment-related, and psychosocial- and finance-related topics using a scale ranging from 0 for “not at all a concern/no discussion” to 100 for “extreme concern/in-depth discussion.” Cronbach α values for concerns about treatment-related issues, the level of discussion about treatment-related topics, and the level of discussion about psychosocial- and finance-related topics were. 81, .95, and .97, respectively.

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Data Analysis

Descriptive statistics and chi-square tests were conducted to examine associations of the sociodemographic characteristics, marital status, and the length of time since initial treatment with the age group. Because of the relatively small sample size, the Mann-Whitney *U* test was performed to examine differences in participants' perceptions about communication with health care providers by age group: younger (<45 years old) and older (≥46 years old) participants. When analyzing missing data, we confirmed that the variables of interest were missing completely at random (Little's missing completely at random test, $P>.05$); thus, the results using the listwise deletion method were reported [13]. All statistical analyses were conducted with Stata 17.0 (StataCorp LLC).

Results

A total of 143 participants completed the survey and provided their year of birth. Among those participants, 140 identified themselves as women and 2 as other. The median age of the participants was 49.0 years with 35% (n=50) aged <45 years and 65% (n=93) aged >46 years. The median age of the younger breast cancer participants was 41.5 years, and that of the older breast cancer participants was 56.0 years. Among all the respondents, 83.6% (n=117) were White with 16.4% (n=23) reporting "other." Latino or Hispanic ethnicity was claimed by 17.0% (n=24). The differences in education levels between the groups were significant and generally high with 18.9% (n=27) having a high school degree or some college education; 48.3% (n=69) held associate or bachelor's degrees, and 32.9% (n=47) mentioned advanced master's, professional, or doctoral degrees. Approximately 78.3% (n=112) of the participants were insured through their employer or had private insurance at the time of their diagnosis. The characteristics of all the participants are presented in Table 1.

Among the set of questions about their concerns regarding treatment-related effects, only 2 responses showed statistically significant differences between the younger (<45 years of age) and the older (>46 years of age) participants, as observed in Table 2. The question about concerns regarding genetic counseling had a mean of 65.40 for all participants with a mean of 73.60 for the younger group and 60.28 for the older group ($P=.04$). The question about concerns related to fertility preservation was significant, with means of 25.60 for all the participants, 45.70 for the younger participants, and 4.59 for older participants ($P=.002$).

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Table 1. Survey participant characteristics.

Characteristics	Total N=143	≤45 years n=50 (35%)	≥46 years n=93 (65%)	Group test *P* value
Age in years, n, Mean (SD)	N/A[b]	50, 40.7 (4.5)	93, 57.6 (9.3)	
Gender, n (%)				N/A
Female	140 (97.9)	50 (100)	90 (96.8)	
Other	2 (1.4)	0 (0)	2 (2.2)	
Missing	1 (0.7)	0 (0)	1 (1.1)	
Race, n (%)				.14
White	116 (81.1)	37 (74)	79 (84.9)	
Other	23 (16.1)	11 (2)	12 (12.9)	
Missing	4 (2.8)	2 (4)	2 (2.2)	
Ethnicity, n (%)				.49
Spanish, Hispanic, or Latino	24 (16.8)	10 (20)	14 (15.1)	
Other	117 (81.8)	40 (80)	77 (82.8)	
Missing	2 (1.4)	0 (0)	2 (2.2)	
Education, n (%)				*.05*
High school graduate or some college with no degree	27 (18.9)	12 (24)	15 (16.1)	
Associate's or bachelor's degree	68 (47.6)	28 (56)	40 (43)	
Master's, professional, or doctoral degree[a]	47 (32.7)	10 (20)	37 (39.8)	
Missing	1 (0.7)	0 (0)	1 (1.1)	
Income in US $, n (%)				.48
<$10,000 to $39,999	26 (18.2)	12 (24)	14 (15.1)	
$40,000 to $79,999	40 (28)	14 (28)	26 (28)	
$80,000 to ≥ $150,000	73 (51)	24 (48)	49 (52.7)	
Missing	4 (2.8)	0 (0)	4 (4.3)	
Marital status, n (%)				.79
Married	94 (65.7)	33 (66)	61 (65.6)	
Not married/other	48 (33.6)	17 (34)	31 (33.3)	
Missing	1 (0.7)	0 (0)	1 (1.1)	
Length of time since initial treatment, n (%)				.37
Still in active treatment	21 (14.7)	10 (20)	11 (11.8)	
Less than 6 months	14 (9.8)	5 (10)	9 (9.7)	
1-4 years	51 (35.7)	18 (36)	33 (35.5)	
More than 5 years	26 (18.2)	6 (12)	20 (21.5)	
Missing	31 (21.7)	11 (22)	20 (21.5)	
Insurance status, n (%)				N/A
Insured through employer or private insurance purchased	112 (78.3)	43 (86)	69 (47.2)	
Medicare or secondary insurance	14 (9.8)	1 (2)	13 (14)	
Medicaid for breast and cervical cancer or Medicaid	3 (2.1)	1 (2)	2 (2.2)	
Military (TRICARE)	4 (2.8)	2 (4)	2 (2.2)	
Not insured	10 (7)	3 (6)	7 (7.5)	

[a]Professional or doctoral degrees: Juris Doctor and Doctor of Medicine.

[b]N/A: Not applicable. Chi-square tests were not possible due to insufficient observations for this category.

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Table 2. Mann-Whitney *U* test results regarding concerns about treatment-related issues (N=143).

From the following list of treatment-related issues, please rate each of the following concerns (0=Does/did not at all concern you; 100=Extreme concern)	Total			≤45 years old			≥46 years old			Group test *P* value[a]
	n	Mean (SD)	Median	n	Mean (SD)	Median	n	Mean (SD)	Median	N/A
Need for genetic counseling	104	65.40 (32.47)	71.00	40	73.60 (30.43)	82.50	64	60.28 (32.88)	59.50	*.04*
Access to fertility preservation	45	25.60 (37.76)	3.00	23	45.70 (43.75)	30.00	22	4.59 (8.77)	1.00	*.002*
Chemotherapy side effects	99	76.32 (29.61)	90.00	39	77.54 (27.69)	82.00	60	75.53 (30.99)	90.00	.77
Mastectomy	109	71.94 (31.78)	80.00	43	77.65 (28.47)	98.00	66	68.23 (33.45)	76.50	.09
Managing all my prescribed medications and treatments	98	64.70 (30.20)	68.00	40	65.05 (24.76)	64.50	58	64.47 (33.66)	72.50	.73
Hair Loss	90	67.08 (32.03)	74.00	37	67.97 (31.68)	75.00	53	66.45 (32.56)	73.00	83
Managing pain and discomfort	104	72.08 (25.05)	76.00	39	75.49 (22.20)	80.00	65	70.03 (26.57)	74.50	.33
Reconstructive surgery	97	74.43 (29.55)	76.00	40	82.23 (21.23)	90.00	57	68.96 (33.30)	78.00	.07
Using medications to manage long-term side effects	97	71.14 (29.06)	85.00	35	69.60 (28.43)	76.00	62	72.02 (29.61)	80.00	.56
Managing ongoing side effects of treatment	107	76.67 (24.97)	84.00	41	76.54 (21.21)	80.00	66	76.76 (27.20)	85.00	.58
Finding undergarments/clothes/wigs to wear after surgery/treatment	93	57.67 (31.71)	60.00	35	57.03 (32.35)	65.00	58	58.05 (31.60)	59.50	.87
Average	N/A[b]	65.73	69.73	N/A	69.85	74.82	N/A	62.31	68.14	N/A
Rollup	N/A	67.99	75.00	N/A	71.06	79.00	N/A	65.98	75.00	N/A

[a]Italicized *P* values are statistically significant.

[b]N/A: not applicable.

The next set of questions focused on transitions to posttreatment care. This was the area where we found the greatest number of significant differences between the younger (<45 years of age) and older participants (>46 years of age), and where the second hypothesis that younger breast cancer survivors experience less support from their health care teams appeared to be best demonstrated. The questions asking about transitions to posttreatment care were ranked based on the extent to which treatment-related topics were discussed by the health care team during the transition to posttreatment care on a scale from 0 indicating "no discussion" to 100 indicating "in-depth discussion." Table 3 provides the results from this set of questions addressing our second hypothesis suggesting that there are areas of communication breakdown or lack of communication between breast cancer survivors and their health care team during transition to posttreatment care. There are "rollups" of the scores in Table 3 and Table 4 that provide a summary of the preaggregated values for the mean and median.

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Table 3. Mann-Whitney *U* test results for posttreatment-related topics (N=143).

To what extent did your health care team discuss the following treatment-related topics with you during your transition from treatment to posttreatment care (0=No discussion; 100=In-depth discussion)?	Total			≤45 years old			≥46 years old			Group test *P* value[a]
	n	Mean (SD)	Median	n	Mean (SD)	Median	n	Mean (SD)	Median	N/A[b]
Which doctor would manage your posttreatment care?	82	56.26 (32.17)	52.00	26	43.96 (32.22)	36.00	56	61.96 (30.78)	59.50	*.02*
When to contact your oncologist vs your primary care doc vs your OB-GYN[c]?	72	46.10 (36.66)	35.00	22	32.77 (37.39)	13.00	50	51.96 (35.12)	50.00	*.03*
What long-term effects of treatment to expect (eg, early menopause)?	77	55.77 (32.31)	60.00	27	43.52 (31.62)	50.00	50	62.38 (31.00)	63.50	*.02*
What should you do for exercise and nutrition?	74	44.45 (29.51)	39.50	23	40.57 (30.18)	35.00	51	46.20 (29.33)	43.00	.45
How frequently you should have follow-up appointments?	85	64.29 (31.23)	68.00	27	54.44 (33.45)	50.00	58	68.88 (29.31)	72.50	*.05*
How often you would need scans/tests?	78	58.35 (31.61)	52.50	24	48.00 (29.00)	48.00	54	62.94 (31.89)	60.50	.06
What are your chances for recurrence/metastatic breast cancer?	82	55.71 (31.84)	55.00	25	47.12 (30.99)	50.00	57	59.47 (31.74)	59.00	.11
What symptoms should you look for recurrence or metastatic breast cancer?	76	50.13 (35.12)	49.50	22	44.10 (34.21)	37.00	54	52.59 (35.50)	51.50	.36
What are your risks for other cancers?	67	47.19 (35.58)	39.00	22	42.27 (37.89)	31.50	45	49.60 (34.58)	49.00	.36
Your survivorship and treatment care plan or next step summary	67	55.91 (34.88)	53.00	19	43.05 (35.73)	45.00	48	61.00 (33.56)	62.50	.06
Average	N/A	53.41	50.35	N/A	53.41	50.35	N/A	57.70	57.10	N/A
Rollup	N/A	53.71	51.00	N/A	44.26	40.00	N/A	57.99	55.00	N/A

[a]Italicized *P* values are statistically significant.

[b]N/A: not applicable.

[c]OB-GYN: obstetrician-gynecologist.

The statistically significant questions where the younger participants responded with lower scores than the older breast cancer survivor participants regarding the extent to which their health care provider discussed transition topics included the following:

- Which doctor (oncologist vs primary care) would manage posttreatment care? The younger participants' mean score was 43.96 vs the older participants' mean score of 61.96 (*P*=.02).
- When to contact your oncologist or primary care doctor or your OB-GYN (obstetrician-gynecologist)? The younger participants' mean score was 32.77, and the older participants' mean score was 51.96 (*P*=.03).
- What long-term effects to anticipate (ie, early menopause)? The younger participants' mean score was 43.52, and that of the older participants was 62.38 (*P*=.02).
- How frequently you should have follow-up appointments? The younger participants' mean score was 54.44, and that of the older participants was 68.88 (*P*=.05).

These results address our second hypothesis and suggest that younger women may have greater needs for patient-centered communication with physicians and health care providers, especially for psychosocial care and during transition to posttreatment care. The results also address our third hypothesis that younger breast cancer survivors need patient-centered communication and information on survivorship and follow-up care during transitions to posttreatment care.

Among the other nonstatistically significant questions, there were several that showed large differences between the 2 means, indicating that these questions too may be important differentiators between the needs of younger and older breast cancer survivors. These included questions about the need for scans/tests or summaries of next treatment steps. Overall, the group mean scores regarding transitions in care were relatively low with an average rollup score of 53.71 for all the questions regarding this stage of care.

Although there were no statistically significant differences in the levels of discussions with health care teams about psychosocial- and finance-related topics between the younger and older participants, the overall scores for the group based on the extent of the discussions with health care providers were all less than 50, ranging from a low mean of 22.54 for concerns

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about remaining medical bills to a high mean of only 34.53 for questions about the need for financial service counseling or support. The rollup of the means and medians for this area of questioning was 30.02. Table 4 shows the means and medians of this group of questions for the entire participant group and for the younger and older groups as well as the average and rollup scores.

Table 4. Scores related to psychosocial and finance-related discussions with health care providers.

To what extent did your health care team discuss the following psychosocial and finance-related topics with you during your transition from treatment to post-treatment care (0=No discussion; 100=In-depth discussion)?	Total			≤45 years old			≥46 years old			Group test *P* value
	n	Mean (SD)	Median	n	Mean (SD)	Median	n	Mean (SD)	Median	N/A[a]
Your concerns about remaining medical bills	48	27.02 (30.65)	16.00	14	18.57 (27.31)	4.00	34	30.50 (31.64)	19.00	.22
Your concerns about cost for posttreatment therapies and medication	44	33.52 (33.01)	28.00	15	21.47 (26.97)	9.00	29	39.76 (34.51)	37.00	.08
Your need for financial service counseling or support	40	34.53 (36.78)	17.00	13	23.15 (27.48)	8.00	27	40.00 (39.81)	20.00	.32
Your need for ongoing emotional/mental support or counseling	48	31.56 (29.75)	26.50	13	23.08 (26.88)	12.00	35	34.71 (30.51)	28.00	.31
Supporting your spouse, children, and family members through posttreatment	32	28.28 (35.41)	8.00	10	15.30 (31.11)	2.00	22	34.18 (36.33)	17.50	.10
Supporting your spouse, children, and family members through a diagnosis of metastatic breast cancer	28	22.54 (31.70)	5.00	9	7.22 (12.74)	0.00	19	29.79 (35.53)	18.00	.06
Average	N/A	29.57	16.75	N/A	18.13	5.83	N/A	34.82	23.25	**N/A**
Rollup	N/A	30.02	17.50	N/A	18.93	5.00	N/A	34.96	25.00	N/A

[a]N/A: not applicable.

Discussion

Principal Findings

Our survey results demonstrated that breast cancer survivors experience barriers or gaps in communication with their health care teams during transition from treatment to posttreatment care. We observed that younger breast cancer survivors have lower statistically significant scores regarding the depth of communications with health care providers pertaining to transitions to posttreatment regarding when to contact which care provider (ie, oncology team vs primary care), what long-term effects to anticipate, and how often they would need follow-up scans or tests. For younger and older participants, the mean scores for what would be considered critically important aspects of cancer survivorship fell below 60 points on the 100-point scale of 0 for “no discussion” and 100 for “in-depth discussion.”

For the questions in the areas of communications with health care team members regarding psychosocial and finance-related topics, our results were comparable to the findings in a nationally representative sample in which limited proportions of cancer survivors reported high-quality discussions with providers after diagnosis, ranging from 29% (n=349) for emotional and social needs to 62% (n=745) for follow-up care recommendations, indicating that 76% experienced suboptimal communication with their cancer care providers [14]. These relatively low scores for patient-provider communication are concerning, especially the apparent lack of discussion about the late and long-term effects of treatment. A number of studies have shown that cancer survivors face many challenging physical and psychological effects of treatment that fundamentally shape their quality of life [14,15]. This concern is well documented, especially for younger survivors [16-23]. Research in this area strongly supports the need for improvement in patient-focused communication among providers and oncology health care team members.

Regarding younger breast cancer survivors as compared to older survivors, patient-specific communication assumes additional importance, as shown by the findings of Champion et al [24]. This work was a retrospective study involving more than 500 breast cancer survivors aged 25 to 50 years, showing that women experienced long-term difficulties with emotional and social functioning, which increased with decreasing age at diagnosis. In their study, younger breast cancer survivors experienced lower vitality and higher rates of depression in comparison to age-matched healthy controls and women who were older at diagnosis. The conclusions drawn by Champion et al suggested that women diagnosed with breast cancer at a younger age (<45) are at significant risk for emotional and psychosocial sequelae during and after breast cancer treatment. Their research suggested that younger women require age-specific psychosocial support, ideally in the context of coordinated multidisciplinary care teams [24,25].

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This need for support is further supported by the study of Johnson et al addressing breast cancer in adolescents and young adults [26]. In this study, the researchers found that concerns about fertility, sexuality, body image, and disruptions in peer and romantic relationships as well as financial and occupational difficulties and fear of death from cancer are more pronounced in younger breast cancer survivors than in older survivors, and that these concerns my contribute to survivor distress [26].

Our study was cross-sectional, thus limiting the ability to draw causal inferences. We could not control for certain variables, such as the cancer site, stage or subtype, provider type, or specialty due to sample size limitations and lack of information. The mix of younger (≤45 years of age) and older (≥46 years of age) participants in our survey, with 35% being younger, is higher than the national ratio of 11% younger (≤45 years of age) patients [3]. This could affect the group means and the rollup scores. This age group is also primarily reflective of Central Texas and especially the Travis County catchment area in which the median age is 34.2 years [27]. However, we may have had responses from other areas of Texas, and thus our survey is not necessarily representative of Central Texas or Texas in general. Our sample was small, partially due to missing data; therefore, this limited our analysis to determining differences in participants' perceptions about communication with health care providers by age group. We confirmed that the missing data met the assumption of MCAR (Little's missing completely at random test, $P>.05$) and employed the listwise deletion method, a common method to generate unbiased and conservative estimates [16].

The survivors' cancer history was self-reported. Our sample was predominantly composed of non-Hispanic Whites and communication differences may exist among patients from diverse racial, ethnic, and cultural backgrounds. There was also the possibility of recall bias, particularly for respondents further from treatment. Our study was conducted during the period of sequester in Central Texas due to the COVID-19 pandemic, which may account for a slightly lower response rate.

Conclusions

Breast cancer survivors' perceptions of conversations with health care professionals revealed missed opportunities for older and younger patients regarding understanding of concerns related to costs, the need for financial services, emotional/mental support counseling, and the need for providing patients' spouses and children with posttreatment support. Participants in this survey emphasized additional support for spouses, children, and family members of those diagnosed with metastatic breast cancer. This research also revealed missed opportunities for enhancing patient-provider communication among younger breast cancer patients during treatment regarding genetic screening and fertility preservation services.

Younger and older breast cancer survivors transitioning from treatment to posttreatment care would benefit from being offered access to psychosocial and financial counseling following breast cancer treatment. These gaps and barriers imply the need for oncology care teams to increase their focus on communications and clarity regarding transitions in care, follow-up care, late or long-term treatment effects, financial support, and psychosocial needs, with special focus on younger breast cancer patients.

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Conflicts of Interest

None declared.

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Abbreviations

BCRC: Breast Cancer Resource Center

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Original Paper

The Use of Telemedicine in Cancer Clinical Trials: Connect-Patient-to-Doctor Prospective Study

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Abstract

Background: Telemedicine is currently being adopted for the management of patients in routine care. However, its use remains limited in the context of clinical trials.

Objective: This study aimed to demonstrate the feasibility of telemonitoring and patient-reported outcomes collection in the context of clinical trials.

Methods: The patients who were included in an interventional oncology clinical trial were eligible. The patients were registered with a digital tool to respond to a patient-reported outcomes questionnaire (ePRO) based on CTCAE (The Common Terminology Criteria for Adverse Events, National Cancer Institute), version 5.0, personalized to their pathology and treatment. An algorithm evaluated the health status of the patient based on the reported adverse events, with a classification in 4 different states (correct, compromise, state to be monitored, or critical state). The main objective was to evaluate the feasibility of remote monitoring via a connected platform of patients included in a clinical trial.

Results: From July 1, 2020, to March 31, 2021, 39 patients were included. The median age was 71 years (range 41-94); 74% (n=29) were male, and 59% (n=23) had metastatic disease. Out of the 969 ePRO questionnaires completed over the course of the study, 77.0% (n=746) were classified as "correct," 10.9% (n=106) as "compromised," and 12.1% (n=117) as "to be monitored" or "critical." The median response time was 7 days (IQR 7-15.5), and 76% (25/33) of the patients were compliant. Out of the 35 patients who answered a satisfaction questionnaire, 95% (n=33) were satisfied or very satisfied with the tool, and 85% (n=30) were satisfied with their relationship with the health care team. There were 5 unscheduled hospitalizations during the study period.

Conclusions: Remote monitoring in clinical trials is feasible, with a high level of patient participation and satisfaction. It benefits patients, but it also ensures the high quality of the trial through the early management of adverse events and better knowledge of the tolerance profile of experimental treatments. This e-technology will likely be deployed more widely in our clinical trials.

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KEYWORDS

telemedicine; clinical trial; neoplasms; patient-reported outcome measures

Introduction

Remote Monitoring of Cancer Patients

Telemedicine has been shown to provide a level of care quality at least equivalent to in-person care, with high levels of patient and health care professional satisfaction [1]. The advantages of remote monitoring of patients are the following: early and real time detection of illnesses, ability to continuously monitor patients, prevention of worsening of illnesses and untimely deaths, cost reduction in hospitalizations, reduction in the number of hospitalizations, more accurate readings without

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interfering with the daily activities of patients, improved efficiency of health care services through the use of digital communication, emergency medical care, service for patients with mobility issues, emergency care for traffic accidents and other injuries, and usage of noninvasive medical interventions [2]. The collection of patient-reported outcomes (PRO) using a telemonitoring approach results in an evaluation closer to the patient's experience of the disease, allowing adjustments to the treatment in order to improve tolerance and compliance. This also improves communication between the health practitioner and the patient [3].

In France alone, 382,000 new cases of cancer are diagnosed every year, and the number of cancer deaths is estimated at 157,400 [4]. Cancer remains a serious public health problem. The use of remote monitoring in the care of cancer patients has shown a significant reduction in mortality compared with standard care [5]. Despite the known benefits, its implementation and use in clinical trials remain limited. Barely 50% of clinical trials in oncology assess the perception of patients, and only 20% of published trials report quality-of-life data and PRO. This figure drops drastically if the study is negative [6,7]. There are various explanations for the lack of such data, including the difficulty in using the existing tools and interpreting their output, as well as the lack of training of the medical teams.

Clinical trials are a critical tool to evaluate new approaches for screening, diagnosis, treatment, and patient care improvements. For drug development, the results of these trials are mandatory for regulatory approval and provide clinicians with new strategies based on efficacy and safety data. Thus, the lack of PRO in the context of clinical trials means that highly relevant information for decision-making is often unavailable to patients, oncologists, and policy makers.

This connect-patient-to-doctor study aimed to demonstrate the feasibility of telemonitoring and PRO collection in the context of clinical trials. It was conducted at the Bégin Military Teaching Hospital, which typically participates in 30 clinical trials every year that include around 50-60 new patients. The primary hypothesis of this paper was that we should see a high level of compliance with the use of the telemonitoring platform, which would thus be a useful complementary tool in the care of the patients, resulting in a better understanding of drug safety.

Methods

This study is a prospective study, conducted in Clinical Research Unit of Bégin Military Teaching Hospital. It was declared to the National Institute for Health (Institut National des Données de Santé, Data MR) and was reported to France's National Commission on Informatics and Liberty, reference 2222625.

Patients

The study ran from July 1, 2020, until March 31, 2021, and included 39 patients. Any patient who was treated at the Bégin Military Teaching Hospital and was included in an interventional oncology clinical trial was eligible for the study. All trials were considered for inclusion, regardless of their phase and promoter type (academic or industrial). There were 2 exclusion criteria for our study: patients who did not agree to use a digital telemonitoring tool and minors (17 years old or less). Patients were included at the time of a hospital visit as long as they were receiving an antitumoral treatment, regardless of the starting date of the clinical trial. Patients with internet access via their smartphone or via a computer were included in the "telemonitoring" cohort. Patients without internet access or with little autonomy from the tool were included in the "call session" cohort and were contacted by telephone at regular intervals. All the patients included in the study signed a consent for this trial.

Study Design

Each cancer patient was allowed to respond to a symptomatology questionnaire personalized to their pathology and treatment. The various symptomatology questionnaires used in the study were created by a pluridisciplinary team of oncologists working with the Cureety team. Each questionnaire includes questions for 10 to 20 adverse events relevant to a specific pathology and treatment. The individual questions follow the CTCAE (Common Terminology Criteria for Adverse Events) grading for each adverse event and mostly use the phrasing of the PRO-CTCAE questions and list of prewritten answers (single-select multiple-choice question); however, they also include some modifications to allow a more objective grading directly by the patient without requiring further evaluation by a health practitioner, making the CTCAE standard usable as part of this digital monitoring tool.

The patients were introduced to and enrolled into the telemonitoring platform by their medical team who also assigned an appropriate questionnaire depending on the patient pathology and treatment. The patients in the telemonitoring cohort were then fully autonomous in the use of the platform, with an initial email that allowed them to create their credentials, followed by an information panel in the web application on their first login; the patients were then free to answer the symptomatology questionnaire as often as they wanted (up to once a day), and would otherwise receive text message reminders every 1 or 2 weeks depending on the questionnaire (see below) with a link to the web application. Patients in the call session cohort were called by the medical team once a week, who went over the questionnaire over the phone if the patient was available and willing to answer. All patients were also free to contact the medical team at any time over the phone or via email. More generally, they were clearly instructed that the telemonitoring tool was not meant to replace more traditional care practices, only to supplement them.

Each reported adverse event (AE) was classified based on CTCAE, version 5.0. For each completed questionnaire, a global health score was computed by an algorithm that weighed the grades of the reported AEs according to their potential severity for the given pathology and treatment. The score was then used to classify the patient into 1 of 4 different states: correct (green), compromised (yellow), to be monitored (orange), or critical (red) (Figure 1).

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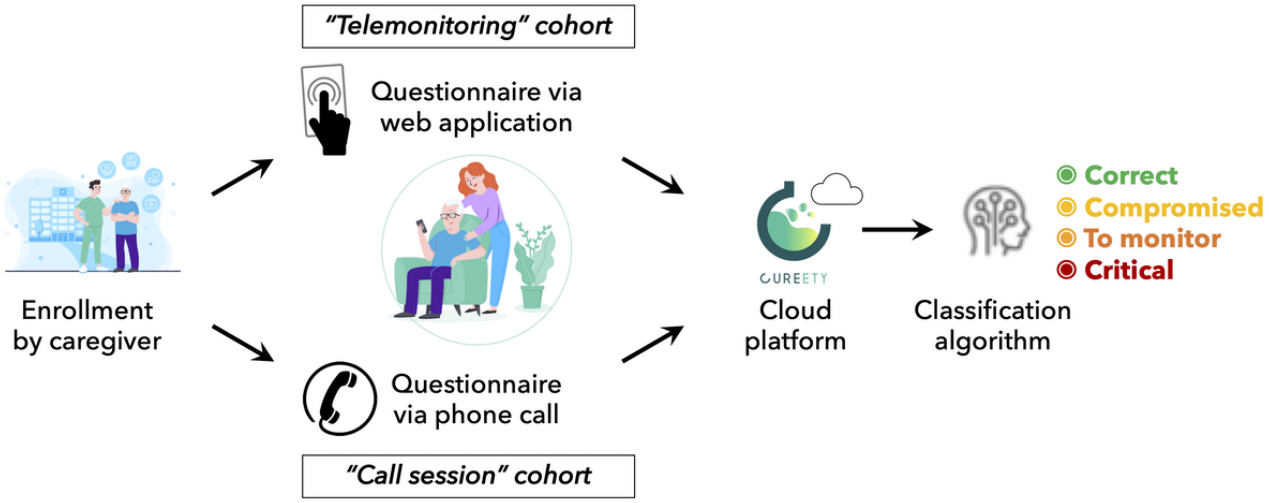
Figure 1. Cancer patient care using the connected telemonitoring platform, Cureety.



At the end of the questionnaire, and for each declared AE, the patient received therapeutic recommendations accordingly. In the case of green or yellow states, the patient received only these therapeutic recommendations. In the case of orange or red states, the patients received the therapeutic recommendations and were also invited to call the hospital or their general practitioner. The clinical research unit's team also received by email an alert for orange and red states. Patients could contact the hospital team at any time if they needed to.

The primary end point was to assess the feasibility of monitoring cancer patients included in clinical trials, using the connected platform. The patients were expected to respond to their questionnaire at least once a week for any treatment that included chemotherapy or immunotherapy, and otherwise every 2 weeks (hormonotherapy, targeted therapy, and radiotherapy, alone or in combination with each other). The patients were thus considered to be compliant if the median frequency of their responses was below this target, with an extra tolerance of 2 days to take into account acceptable compliance gaps. Compliance was only assessed for patients who were monitored for at least 30 days to ensure enough data had been collected to calculate the frequency of their reports.

The secondary end point was to assess the patient's tolerance profile during the study, the patient's satisfaction, and the number of unscheduled hospitalizations. To evaluate satisfaction, at the end of the study, all patients had to complete a satisfaction questionnaire, which contained 8 questions with a 5-level Likert scale for the responses: "strongly disagree," "disagree," "neither agree nor disagree," "agree," and "strongly agree." A final open-ended question also allowed the patient to leave additional comments and provide suggestions about the platform.

Data Collection and Measurements

We collected demographic data (age at inclusion, sex, and comorbidities), disease characteristics (primitive, histology, and stage at inclusion), phase of clinical trial, and type of treatment received. The individual AEs, the grades reported by the patients, as well as the global health status were collected throughout the duration of the clinical trials in which the patients participated. The number of unscheduled hospitalizations was collected from the patient medical records.

Statistical Analysis

The PRO data (AEs and health status) were collected digitally and entered directly into the Cureety platform database for both the "telemonitoring" cohort (via the patient application), or the "call session" cohort (via the caregiver application, while on the phone with the patient), eschewing the need for a paper questionnaire and ensuring higher data integrity. The entirety of the digital tool including the web application, the cloud server collecting the data, and the classification algorithm running on that server are developed and managed by the Cureety company. Because it hosts health data of patients in France, the entirety of the technical stack is compliant with the "Hébergeur de données de santé" (health data storage) regulation, which encompasses the ISO (international information security standard) 27001 norm, together with additional rules, and ensures stringent security constraints are in place to protect the patient data. To access the platform, the patients had to create an account and use a username and password combination.

The data were then extracted, analyzed, and formatted using Python (Python Software Foundation) scripts. For descriptive data, median and interquartile range (minimum and maximum) were also indicated.

Results

Characteristics of the Patients Included in the Study

A total of 39 patients were included in our study between July 1, 2020, and March 31, 2021, including 9 in the call session cohort (Figure 2). The median age was 71 years (range 41-94), 74% (n=29) were male, and 69.2% (n=27) presented at least one comorbidity. There was a broad range of primary tumors including prostate cancer (n=23), lung cancer (n=12), breast cancer (n=3), and bladder cancer (n=1). Moreover, 59% (n=23) of the patients had a metastatic disease.

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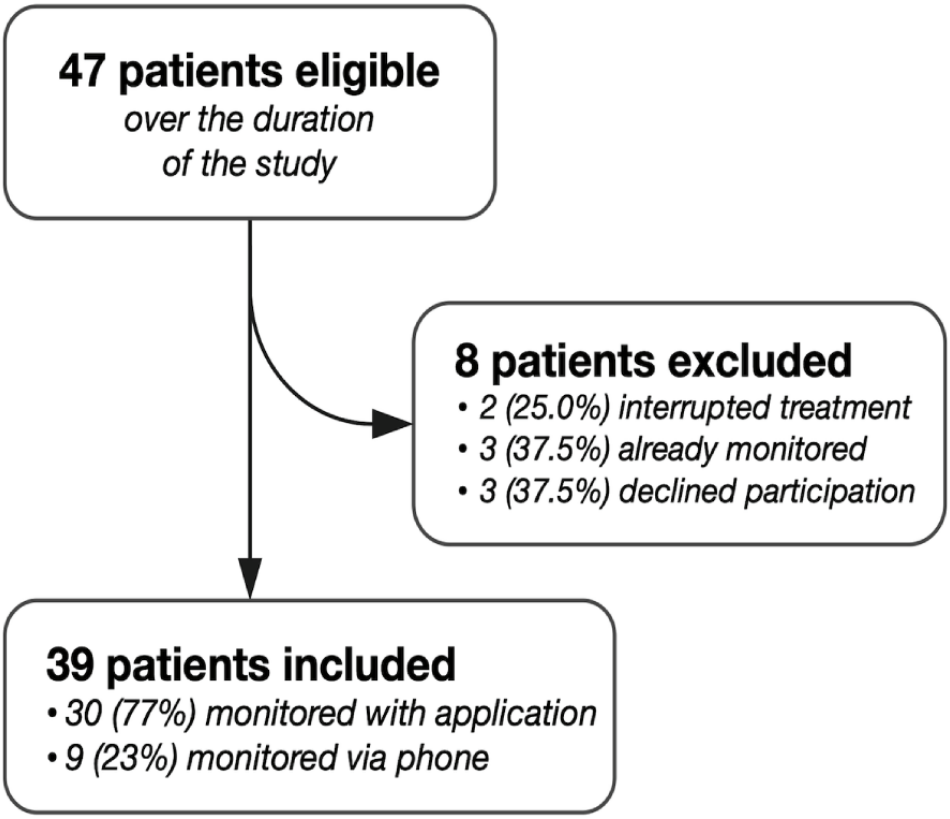
Figure 2. Study flow diagram.



The patients were included in clinical trials in phase III (30, 76.9%), phase II/III (3, 7.7%), phase I (3/39, 7.7%), phase II (2/39, 5.1%) and phase I/II (1/39, 2.6%). There was a broad range of treatment, primarily chemotherapy alone (3/39, 7.7%), chemotherapy with immunotherapy (5/39, 12.8%), new generation of hormonotherapy (11/39, 28.2%), immunotherapy with targeted therapy including PARP (poly adenosine diphosphate-ribose polymerase) inhibitors (3/39, 7.7%), and immunotherapy alone (1/39, 2.6%). Baseline characteristics are summarized in Table 1.

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Table 1. Baseline characteristics of patients.

Variables	Values
Median age (years), range	71, 41-94
Patients, N (%)	39 (100)
Gender, n (%)	
Female	10 (25.6)
Male	29 (74.4)
Comorbidities, n (%)	
Cardiovascular	13 (33.3)
Renal failure	1 (2.6)
Chronic obstructive pulmonary disease	1 (2.5)
Others	12 (30.8)
None	12 (30.8)
Location of cancer, n (%)	
Prostate	23 (59)
Lung	12 (30.7)
Breast	3 (7.7)
Bladder	1 (2.6)
Stage, n (%)	
Metastatic	23 (59)
Localized	12 (30.8)
Localized advanced	2 (5.1)
Oligometastatic	2 (5.1)
Clinical trial phase, n (%)	
I	3 (7.7)
I/II	1 (2.6)
II	2 (5.1)
II/III	3 (7.7)
III	30 (76.9)
Type of treatment, n (%)	
Chemotherapy and immunotherapy	5 (12.8)
Chemotherapy	3 (7.7)
New generation of hormonotherapy	11 (28.2)
Hormonotherapy and radiotherapy	4 (10.2)
Hormonotherapy and targeted therapy	3 (7.7)
Immunotherapy	1 (2.6)
Immunotherapy and targeted therapy	3 (7.7)
Targeted therapy	3 (7.7)
Conjugated antibody	1 (2.6)
Chemotherapy and hormonotherapy	2 (5.1)
Other: adapted physical activity	3 (7.7)

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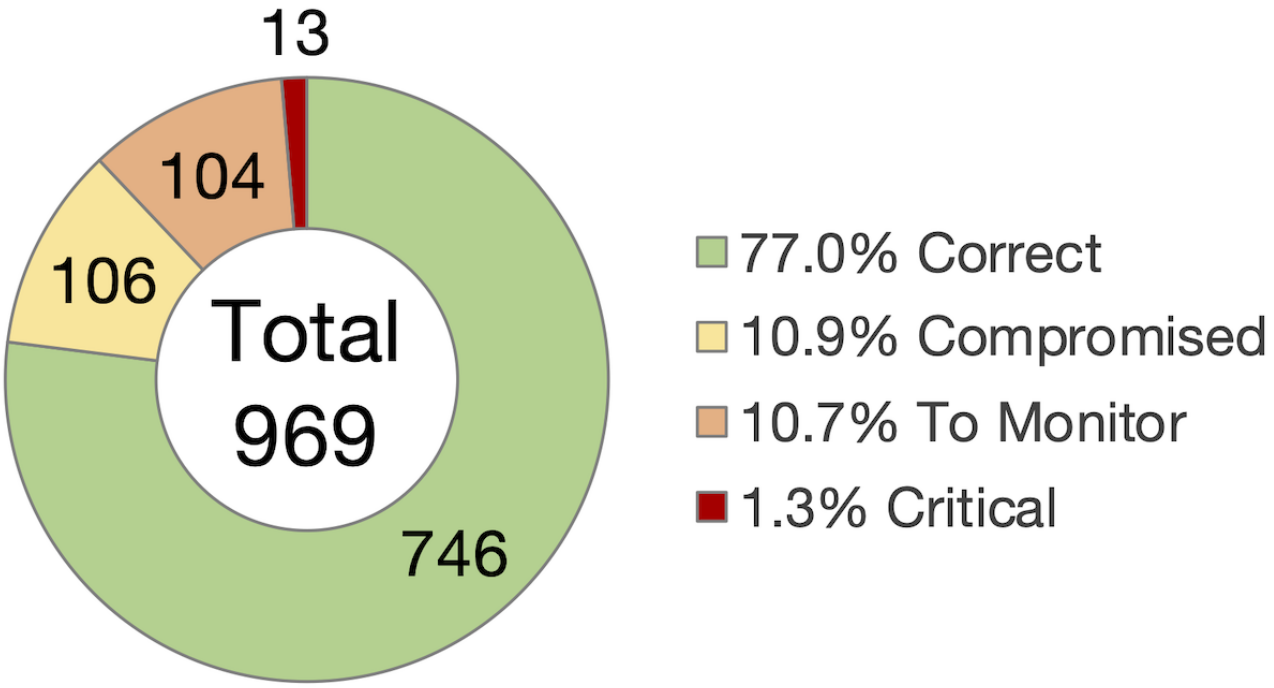
Patient-Reported Outcomes on Adverse Events

Out of the 969 ePRO (patient-reported outcomes questionnaires) completed by the patients, 77.0% (n=746) corresponded to a "correct" state, 10.9% (n=106) to a "compromised" state, 10.7% (n=104) to a state "to be monitored," and 1.3% to a "critical" state (n=13), as shown in Figure 3. These questionnaires correspond to 15,042 AE questions answered, among which there were 84 (0.56%) AEs of grade 3 reported and 37 (0.25%) AEs of grade 4.

Figure 3. Distribution of health status for the 969 questionnaires completed by the patients over the duration of the study.



Among the 39 patients, the median response was 7 days (IQR 6.25-8.75) for patients whose target compliance was 7 days (chemotherapy or immunotherapy), and 7 days (IQR 7-16) for patients whose target compliance was 14 days (other types of treatment). Compliance was calculated for the 33 applicable patients (the other 6 patients were excluded because their participation was shorter than 30 days) and was found to be 75.8% (n=25). Of the 25 compliant patients, 92% (n=23) were still enrolled in their respective clinical trials at the end of the analysis period (March 31, 2021).

In the group with hormonotherapy, 71% (10/14) of the patients were compliant. The global tolerance of these patients was good at 92% (fraction of time where the health state was green or yellow). In the group with targeted therapy, 75% (3/4) of the patients were compliant, with a good global tolerance of 93%. In the group with combined therapies, 77% (10/13) of the patients were compliant (n=10 out of 13) with a good global tolerance of 74%.

Six patients stopped their clinical trial because of death or disease progression. Moreover, 5 unscheduled hospitalizations were recorded during the course of this study, 2 related to AEs and 3 due to disease progression (1 is not represented in Figure 4, as it happened after the end of the analyzed timeline) (Figure 4).

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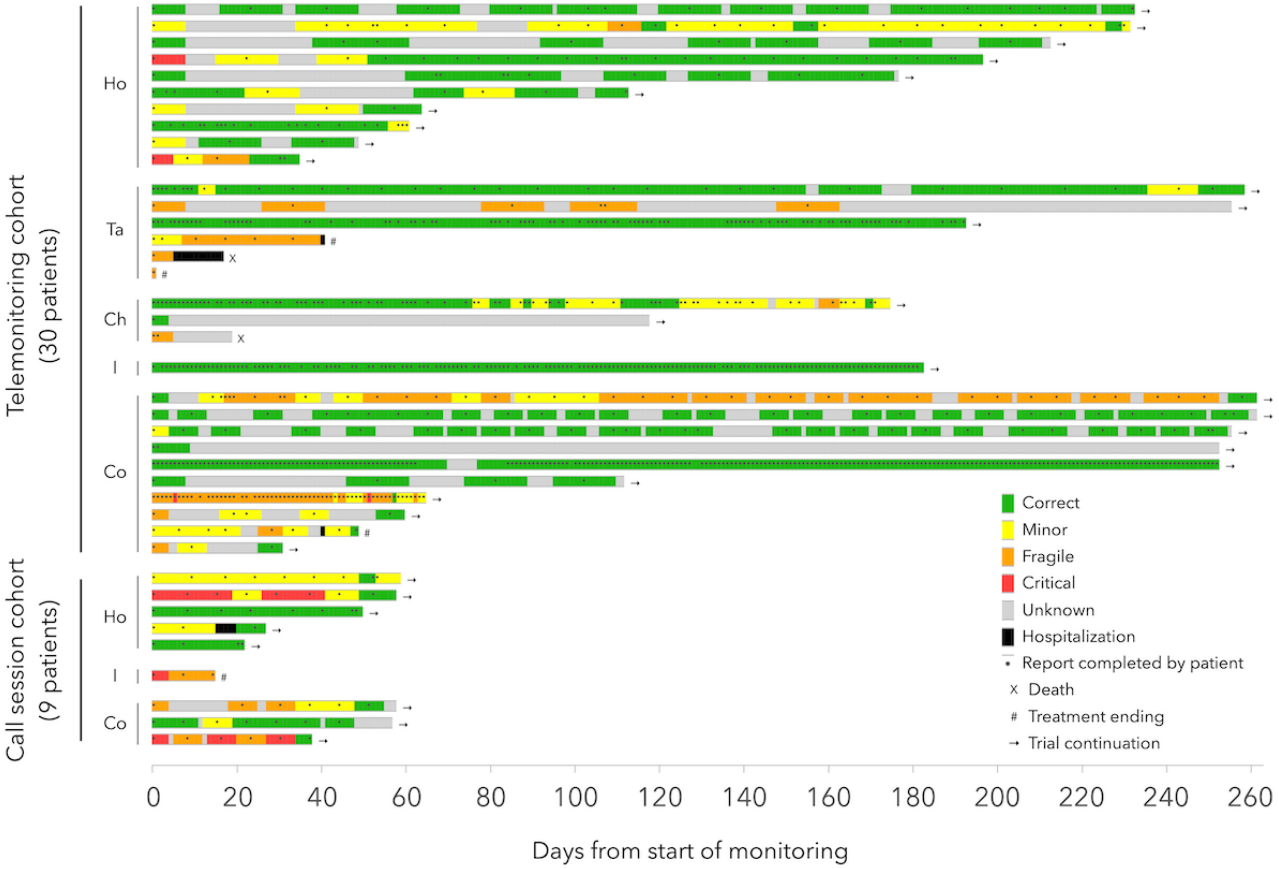
Figure 4. Timelines showing the health status (from the patient-reported outcomes questionnaires) and clinical events of each patient. Ch: chemotherapy; Co: combined therapy; Ho: hormonotherapy; I: immunotherapy; Ta: targeted therapy.



Satisfaction

When prompted, 35 patients completed the satisfaction questionnaire (Figure 5), including all the patients in the "call session" cohort (9/35, 26%). The answers show that 94% (n=33) were satisfied with the monitoring platform, including 51% (n=17) who were very satisfied, and 54% (n=19) estimated that this tool improved the management of their AEs. Additionally, 85% (n=30) of the patients were satisfied with their relationship with their health care team, particularly via the platform, including 66% (n=20) who were very satisfied.

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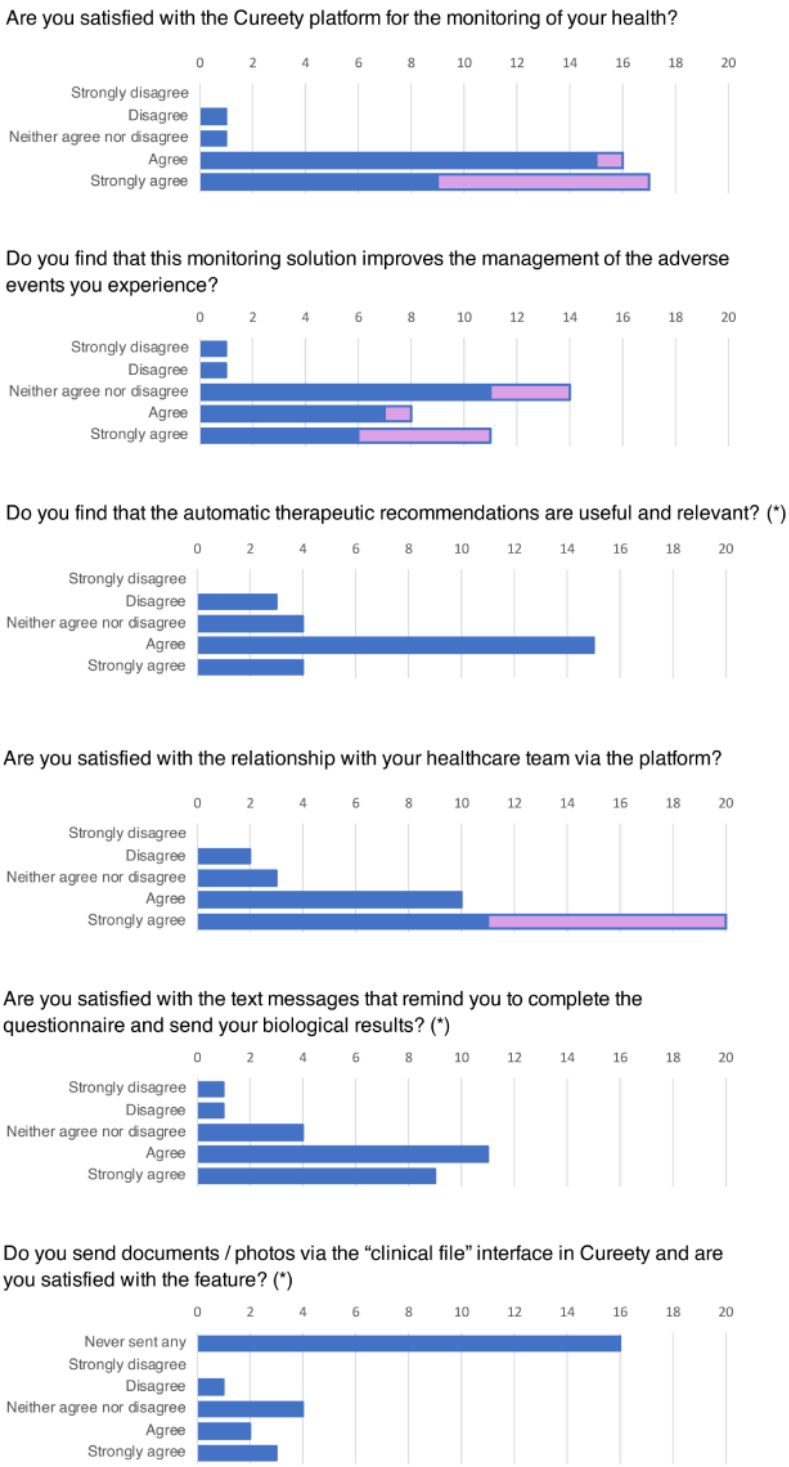
Figure 5. Satisfaction questionnaire results. Asterisks (*) indicate the question was only asked to patients in the telemonitoring cohort.



Discussion

Principal Findings

This connect-patient-to-doctor study is to our knowledge the first study evaluating, via a connected platform, the remote monitoring of cancer patients who are included in clinical trials.

Remote monitoring has already been shown to benefit the management of patients with chronic pathologies such as diabetes, psychiatric and cardiovascular diseases, as well as cancer [8-11]. The benefit is not just at the clinical level but is also medico-economic [11,12]. Kim et al [11] evaluated the impact of remote monitoring versus standard care in the management of patients with type 2 diabetes. In this meta-analysis, 6855 patients were included. Telemonitoring was associated with a significant decrease in glycated hemoglobin levels compared with usual care (weighted mean difference -0.42%, 95% CI -0.56 to -0.27).

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In addition, telemedicine reduces geographic inequalities in access to care. Russo et al [12] reported a benefit of telemedicine on travel time savings as well as travel costs. They noted a gain of 145 miles and 142 minutes per trip with an average savings of US $18,555 per year.

The implementation of telemedicine in our current practice is favored by the increase in the use of connected objects, with more than 90% of patients having a cell phone and 87% using the internet [13].

Telemedicine, especially telemonitoring, provides direct information on the patient's tolerance of the treatment. This practice addresses the discrepancy in AE grading when comparing perception by the patient and interpretation by the care team [14]. It also has therapeutic and psychological benefits for the patient as well as on treatment adherence [15].

Bash et al [16] evaluated the impact of symptom monitoring in the management of 766 cancer patients and found a significant improvement in the patients' quality of life from the monitoring (34% versus 18%, $P<.001$). Remote monitoring was also found to improve the overall survival of patients. Denis et al [17] evaluated the impact of a remote monitoring platform on the overall survival of patients with bronchial cancer, compared with the standard practice. The study showed a 68% reduction in mortality risk in patients benefiting from the remote monitoring platform (hazard ratio 0.32, 95% CI 0.15-0.67, one-sided $P=.002$).

A closer monitoring of the tolerance to treatments thus allows a better management of the patients and has a proven impact on their quality of life. Here, we demonstrated the feasibility of remote monitoring for patients included in clinical trials, with a 76% compliance rate and a high satisfaction rate (94%), all without interfering with the ongoing clinical trials. The patients continued to perform the actions required by their respective trials, in addition to the reporting of AEs via the digital platform.

Our telemonitoring platform allowed us to determine the treatment tolerance profile for each patient during the entire study. It provided therapeutic advice adapted to the grade of the reported AEs. The number of unscheduled hospitalizations observed during the study period (n=5) appeared lower than what the medical team had observed in prior years (22 during the same period of time in 2020), suggesting that telemonitoring may have a positive impact on patient management. We will need a larger dedicated study to properly determine the impact of telemonitoring on unscheduled hospitalizations.

In fact, such an impact was shown by the CAPRI (Cancérologie parcours région Ile de France [Oncology Pathway in the Ile de France Region]) study [18]. This randomized study included 609 cancer patients receiving oral therapy and compared the use of a mobile telemedicine application, combined with follow-up by nurses, with standard care. The study showed a significant decrease in unscheduled hospitalizations, at 15.1% versus 22% ($P=.04$).

Our algorithm shows the accumulated impact of each AE, weighed by their grade level, instead of just considering them independently of each other, thus better reflecting the overall state of the patients. For each clinical trial, we could then estimate the tolerance profile of the patients, as measured by the percentage of time when the health state was "green" or "yellow." The tolerance levels were good: at 92% for patients receiving hormonotherapy, at 93% for targeted therapies, and at 74% for combined therapies.

Postel-Vinay et al [19] reported the importance of long-term monitoring of the tolerance of treatments, starting from phase I in order to better determine the recommended dose for the later trial phases. In addition, the AEs and their grades seem to be predictive factors of the treatment efficacy. Socinski et al [20] recently evaluated immune-related adverse events (irAEs) in a pooled analysis treatment in patients with metastatic non–small cell lung cancer receiving chemotherapy with or without immunotherapy. They reported that patients who experienced an irAE had a gain in overall survival compared to those who did not (hazard ratio 0.69, 95% CI 0.60-0.78). This gain was mostly for patients with grade 1-2 irAE, as compared to patients with grade 3-4, with a median overall survival of 33 months (hazard ratio 0.72, 95% CI 0.59-0.89) and 29.9 months (hazard ratio 0.87, 95% CI 0.61-1.25), respectively.

The responses to the satisfaction questionnaire demonstrated a high level of satisfaction with the platform. Of the 26 patients who used the application for remote monitoring and who responded, 58% were satisfied, and 35% were very satisfied. Of the 9 patients in phone call sessions, 89% were very satisfied. The patients also had a very favorable opinion of the patient-care team relationship, with 86% of the patients being satisfied. This shows that the bond between the patients and their health care team was maintained, allowing for increased compliance and continuation of the clinical trials.

The remote monitoring approach also has an impact on the clinical trial data. The recurrent reporting by the patients provides a more accurate, more complete, and more frequent view of the treatment tolerance under investigation. This information is essential for the evaluation and approval of experimental treatment and is an important complement to the efficacy data.

Digital remote monitoring limits data loss and increases the clarity and accuracy of safety data during clinical trials. With a traditional approach, the reporting of AEs during a clinical trial is often incomplete or missing and is delayed. Allen et al [21] reported the limitations of the current AE reporting methods, with investigator-patient discrepancy and biases introduced by patient memory limitation.

Remote monitoring also allows to limit patient traveling to the care center, while ensuring the smooth running of the trial. Repeated travel is often a source of discontinuation of trial participation and an obstacle to patient enrollment. By reducing the need for in-person visits, remote monitoring helps reduce the duration of a trial, accelerate the collection of the results, and reduce costs [13].

The COVID-19 pandemic has strongly impacted our health system and has resulted in the interruption of many trials during the first wave. The use of e-technologies at each stage of clinical trials during such events is a clear way to modernize clinical trials and lift the obstacles that slow their implementation. These

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technologies can ensure remote trial approval and initiation, remote monitoring, remote visits, and the treatment of participants [22]. Of note, the entirety of the study was conducted during periods of high COVID-19 prevalence in France. Despite the pandemic, compliance with the use of the telemonitoring tool was high, which is very encouraging. By potentially reducing the risk of hospitalizations, such tools protect the patient from exposure to COVID-19 at the hospital.

These encouraging results should now be validated on a larger cohort with patients in clinical trials.

Conclusion

Remote monitoring in clinical trials is feasible, with a high level of patient participation and satisfaction. It not only benefits patients, but also ensures the high quality of the trial, through the early management of adverse events, better knowledge of the tolerance profile of experimental treatments, and the removal of several biases that typically affect such trials. This e-technology should be deployed routinely as part of our daily practice and in our clinical trials.

Conflicts of Interest

CP is an employee and own shares of Cureety.

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Abbreviations

AE: adverse event
CAPRI: Cancérologie parcours région Ile de France
CTCAE: Common Terminology Criteria for Adverse Events
ePRO: patient-reported outcomes questionnaire
irAE: immune-related adverse event
ISO: international information security standard
PARP: poly adenosine diphosphate-ribose polymerase
PRO: patient-reported outcomes

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Original Paper

Twitter Conversations About Pancreatic Cancer by Health Care Providers and the General Public: Thematic Analysis

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Abstract

Background: There is a growing interest in the pattern of consumption of health-related information on social media platforms.

Objective: We evaluated the content of discussions around pancreatic cancer on Twitter to identify subtopics of greatest interest to health care providers and the general public.

Methods: We used an online analytical tool (Creation Pinpoint) to quantify Twitter mentions (tweets and retweets) related to pancreatic cancer between January 2018 and December 2019. Keywords, hashtags, word combinations, and phrases were used to identify mentions. Health care provider profiles were identified using machine learning and then verified by a human analyst. Remaining user profiles were classified as belonging to the general public. Data from conversations were stratified qualitatively into 5 domains: (1) prevention, (2) survivorship, (3) treatment, (4) research, and (5) policy. We compared the themes of conversations initiated by health care providers and the general public and analyzed the impact of the Pancreatic Cancer Awareness Month and announcements by public figures of pancreatic cancer diagnoses on the overall volume of conversations.

Results: Out of 1,258,028 mentions of pancreatic cancer, 313,668 unique mentions were classified into the 5 domains. We found that health care providers most commonly discussed pancreatic cancer research (10,640/27,031 mentions, 39.4%), while the general public most commonly discussed treatment (154,484/307,449 mentions, 50.2%). Health care providers were found to be more likely to initiate conversations related to research (odds ratio [OR] 1.75, 95% CI 1.70-1.79, $P<.001$) and prevention (OR 1.49, 95% CI 1.41-1.57, $P<.001$) whereas the general public took the lead in the domains of treatment (OR 1.63, 95% CI 1.58-1.69, $P<.001$) and survivorship (OR 1.17, 95% CI 1.13-1.21, $P<.001$). Pancreatic Cancer Awareness Month did not increase the number of mentions by health care providers in any of the 5 domains, but general public mentions increased temporarily in all domains except prevention and policy. Health care provider mentions did not increase with announcements by public figures of pancreatic cancer diagnoses. After Alex Trebek, host of the television show *Jeopardy*, received his diagnosis, general public mentions of survivorship increased, while Justice Ruth Bader Ginsburg's diagnosis increased conversations on treatment.

Conclusions: Health care provider conversations on Twitter are not aligned with the general public. Pancreatic Cancer Awareness Month temporarily increased general public conversations about treatment, research, and survivorship, but not prevention or policy. Future studies are needed to understand how conversations on social media platforms can be leveraged to increase health care awareness among the general public.

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KEYWORDS

pancreatic cancer; Twitter; general public; health care providers; social media; Creation Pinpoint; thematic analysis

Introduction

Social media platforms have emerged as tools for patients to access general health-related information and stay up-to-date with the latest therapeutic advancements [1,2]. Social media allows sharing information on cancer screening, prevention, treatment, and survivorship [3-6]. Apart from patients with cancer and their caregivers, cancer centers and patient advocacy groups use social media to disseminate content for patient education and fundraising activities [7]. There is a growing interest in the pattern and nature of the consumption of information by the general public through these platforms. Twitter is a micro-blogging website that can be used for sharing content with users around the world in real time. Tweets (short messages that are limited to a maximum of 280 characters) serve as a quick and efficient source of information that can then be liked, shared (retweeted) or commented on by other users to amplify and to maximize outreach on a common platform [8].

Pancreatic cancer is an intractable malignancy that is associated with a heavy burden of symptoms and poor overall survival [9]. Patients, caregivers, care teams, and researchers use Twitter as a platform to connect and share information related to pancreatic cancer treatments. It has also been used as a platform for advocating for needs and concerns that are unique to patients with pancreatic cancer [10]. However, there is a need to further analyze factors that drive these conversations and how they can be used as opportunities for initiating discussions on topics such as early detection, policy reforms, and survivorship. Additionally, several high-profile public figures have developed pancreatic cancer in recent years. Studying the impact of these events on the volume and nature of conversations can serve as a valuable case study in evaluating the influence of social media on cancer awareness.

We conducted the current analysis to study the themes and dynamics of conversations around pancreatic cancer on Twitter. We looked to study how health care providers and the general public use this platform. We also investigated the impact of Pancreatic Cancer Awareness Month and the diagnoses of public figures on conversations about pancreatic cancer.

Methods

We used an online analytical tool (Creation Pinpoint) to quantify Twitter mentions (tweets and retweets) related to pancreatic cancer made between January 2018 and December 2019. Keywords, hashtags, word combinations, and phrases were used to search for Twitter mentions related to pancreatic cancer. Perspectives from Twitter users were then distilled based on their online behaviors. Machine learning techniques were used to identify health care providers based on their Twitter profile description (commonly known as a Twitter bio). All health care provider profiles were then verified by a human analyst based on professional websites and other sources. Duplicate profiles or profiles that could not be verified were excluded. Only physicians were included as health care providers. In the final analysis, 13,788 health care providers were included. Analyst decisions were verified in a quality check performed by a data quality supervisor (Figure 1). All remaining user profiles were classified as belonging to the general public. After identification of tweets related to pancreatic cancer, data from conversations were analyzed and stratified qualitatively using keywords, combinations, and phrases into 5 domains (Table 1).

The month of November is Pancreatic Cancer Awareness Month. We analyzed the effect of Pancreatic Cancer Awareness Month in 2018 and 2019 on Twitter mentions in each of the 5 domains. Two prominent personalities announced a diagnosis of pancreatic cancer during the study period: Alex Trebek, host of the television show *Jeopardy*, in March 2018 and Justice Ruth Bader Ginsburg in August 2019. Additionally, Aretha Franklin passed away from pancreatic cancer in August 2018. We studied the effect of these 3 public figure cancer diagnoses on Twitter conversations initiated by health care providers and the general public in the domains described above.

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Figure 1. The analytical tool used in this study, Creation Pinpoint, uses machine learning algorithms to identify possible health care provider profiles on Twitter that are later confirmed and verified by data analysts.

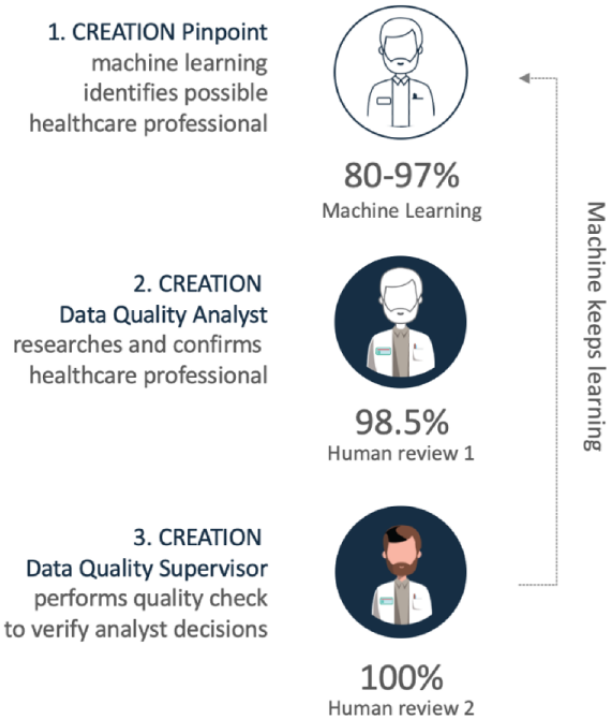

Table 1. Search strategies for identification of tweets related to pancreatic cancer and further categorization into 5 domains: prevention, survivorship, treatment, research, and policy.

Search term	Keywords, combinations, and phrases
Pancreatic cancer	(
	pancchat OR pancan OR pancreaticcancer* OR pancreascancer* OR WorldPancreaticCancerDay OR #WPCD
	OR ((pancreatic OR Pancreas OR pancpath OR acinar OR vipoma OR somatostatinoma OR glucagonoma OR insulinoma OR gastrinoma OR pseudopapillary) AND (cancer OR adenocarcinoma OR carcinoma OR malignant OR tumor OR tumour))
	OR ((PDAC OR PancNET OR PNET) AND (pancreatic OR Pancreas OR pancpath OR cancer OR tumor OR tumour OR mutated OR metastatic))
	OR ((pancreatic OR Pancreas OR pancpath OR acinar OR vipoma OR somatostatinoma OR glucagonoma OR insulinoma OR gastrinoma OR pseudopapillary OR PDAC OR PancNET OR PNET) AND (gemcitabine OR paclitaxel OR FOLFIRI OR mFOLFIRI OR FOLFOX OR fluorouracil OR 5FU OR irintecan OR irinotecan OR everolimus OR evorolimus OR oxaliplatin OR cisplatin OR “demplatin pegraglumer” OR capacitabine OR capecitabine OR docetaxel OR carboplatin OR glufosfamide OR glucosphamide OR leucovorin OR folinic OR tetrahydrofolic OR pembralizumab OR pembrolizumab OR nivolumab OR ipulimumab OR ipalimumab OR ipilimumab OR ipilumab OR cabiralizumab OR cabaralizumab OR cabarilizumab OR urelumab OR olaratumab OR talabostat OR cobimetanib OR cobimetinib OR anetumab OR epacadostat OR atezolizumab OR pamrevlumab OR pegilodecakin OR PEGylated OR PEGPH20 OR pegvorhyaluronidase OR avelumab OR bempegaldesleukin OR erlotinib OR sunitinib OR olaparib OR rucaparib OR napabucasin OR masican OR masitinib OR velaparib OR veliparib OR GVAX OR CRS207 OR Gemzar OR infugem OR onxol OR taxol OR abraxane OR adrucil OR efudex OR efudix OR carac OR onivyde OR campto OR afinitor OR zortress OR eloxatin OR platinol))
	)
	AND site:twitter.com
Prevention	prevent* OR screen OR screening OR ((reduce OR decrease OR lower OR limit) NEAR/3 (risk))
Survivorship	survivor* OR survival OR “OS” OR “PFS” OR overcome OR beat
Treatment	treat OR treatment* OR treating OR gemcitabine OR paclitaxel OR “nab-paclitaxel” OR FOLFIRI* OR mFOLFIRI*,…….
Research	Research OR study OR trial OR trials OR studies OR data
Policy	policy OR policymak* OR ((NIH or NCI)) AND (fund*)) OR (insurance AND expan*)

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Results

Classification by Domain

We identified a total of 1,258,028 English-language mentions related to pancreatic cancer from January 2018 to December 2019, out of which 62,439 were from health care providers and 1,195,598 were from the general public. Out of 1,258,028 mentions, we identified a total of 313,668 unique mentions (27,031 by health care providers and 307,449 by the general public) that were classified into the 5 domains of prevention, treatment, research, survivorship, and policy. Health care providers most often discussed pancreatic cancer research (10,640/27,031 mentions, 39.4%) while the general public most often discussed treatment (154,484/307,449 mentions, 50.2%). Health care providers focused the least on policy (28/27,031 mentions, 0.1%); the general public also focused the least on policy (93/27,031 mentions, 3.3%). A comparative analysis showed that health care providers were more likely to initiate conversations related to research (odds ratio [OR] 1.75, 95% CI 1.70-1.79, $P<.001$) and prevention (OR 1.49, 95% CI 1.41-1.57, $P<.001$) whereas the general public took the lead in the domains of treatment (OR 1.63, 95% CI 1.58-1.69, $P<.001$) and survivorship (OR 1.17, 95% CI 1.13-1.21, $P<.001$). As shown in Figure 2, health care providers were not found to be more likely to initiate conversations in the domain of policy when compared to the general public (OR 0.82, 95% CI 0.55-1.21, $P=.32$). The temporal distribution of mentions in each category for both health care providers and the general public is shown in Table 2.

Figure 2. Forest plot depicting the odds ratio for conversations related to pancreatic cancer initiated by health care providers and the general public in the domains of policy, research, treatment, survivorship, and prevention.

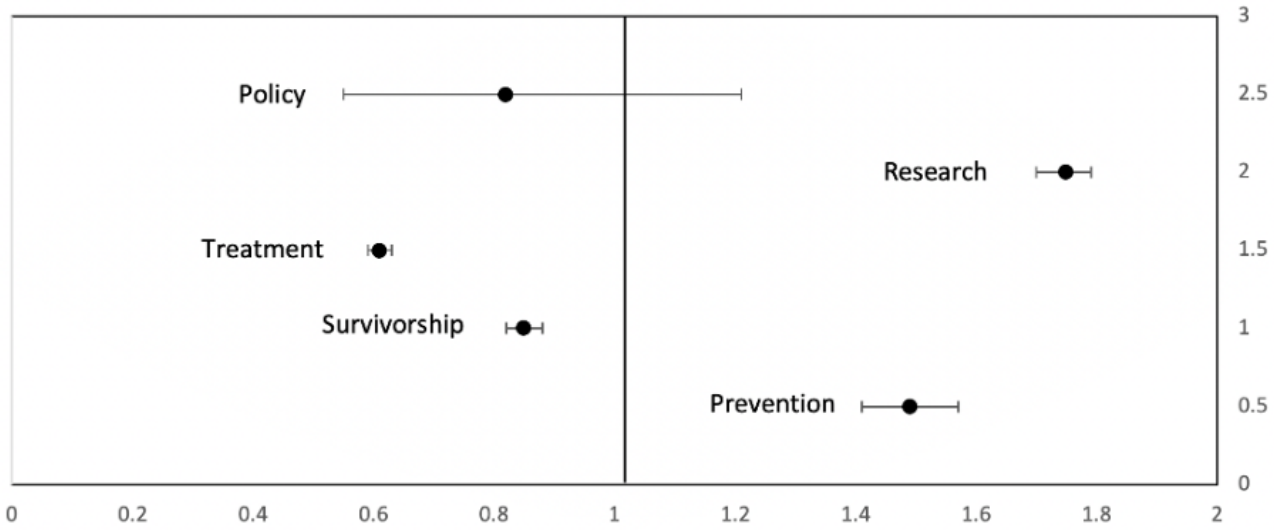

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Table 2. Stratification of Twitter mentions by health care providers and the general public from January 2018 to December 2019.

Month, year	Prevention, n (%)		Survivorship, n (%)		Treatment, n (%)		Research, n (%)		Policy, n (%)	
	HCP[a]	GP[b]	HCP	GP	HCP	GP	HCP	GP	HCP	GP
Jan 2018	62 (8.3)	620 (10.2)	132 (5.9)	1276 (4.6)	435 (9.5)	3409 (6.7)	405 (8.3)	4005 (10.0)	0 (0)	2 (0.8)
Feb 2018	89 (11.9)	648 (10.6)	97 (4.4)	967 (3.5)	184 (4.0)	2076 (4.1)	314 (6.4)	2907 (7.3)	0 (0)	2 (0.8)
Mar 2018	76 (10.2)	547 (8.9)	98 (4.4)	2805 (10.1)	347 (7.6)	3498 (6.9)	372 (7.6)	3151 (7.9)	2 (18.2)	37 (14.2)
April 2018	69 (9.2)	377 (6.2)	115 (5.2)	1026 (3.7)	285 (6.2)	2050 (4.0)	410 (8.4)	2521 (6.3)	1 (9.1)	16 (6.2)
May 2018	65 (8.7)	649 (10.7)	158 (7.1)	1576 (5.7)	438 (9.6)	4841 (9.5)	424 (8.7)	3162 (7.9)	1 (9.1)	17 (6.5)
June 2018	54 (7.3)	286 (4.7)	531 (23.9)	3510 (12.6)	903 (19.7)	5092 (10.0)	854 (17.5)	6322 (15.8)	1 (9.1)	12 (4.6)
July 2018	35 (4.7)	248 (4.1)	95 (4.3)	1073 (3.9)	244 (5.3)	2811 (5.5)	255 (5.2)	2629 (6.6)	0 (0)	3 (1.2)
Aug 2018	47 (6.3)	634 (10.4)	137 (6.2)	3360 (12.1)	244 (5.3)	4184 (8.2)	380 (7.8)	3285 (8.2)	1 (9.1)	6 (2.3)
Sept 2018	36 (4.8)	250 (4.1)	122 (5.5)	1046 (3.8)	246 (5.4)	2441 (4.8)	392 (8.0)	2780 (6.9)	0 (0)	1 (0.4)
Oct 2018	80 (10.7)	833 (13.7)	114 (5.1)	1242 (4.5)	231 (5.0)	3816 (7.5)	287 (5.9)	2497 (6.2)	1 (9.1)	108 (41.5)
Nov 2018	106 (14.2)	802 (13.2)	297 (13.4)	4137 (14.9)	552 (12.04)	13,428 (26.5)	460 (9.4)	4907 (12.3)	3 (27.3)	52 (20.0)
Dec 2018	29 (3.9)	201 (3.3)	321 (14.5)	5796 (20.8)	472 (10.3)	3106 (6.1)	331 (6.8)	1793 (4.5)	1 (9.1)	4 (1.5)
Jan 2019	60 (6.2)	390 (4.7)	178 (7.8)	1350 (3.9)	337 (3.0)	3365 (3.0)	506 (8.8)	2844 (5.8)	0 (0)	1 (0.4)
Feb 2019	74 (7.6)	534 (6.4)	124 (5.4)	1543 (4.5)	212 (3.8)	3200 (2.9)	357 (6.2)	3109 (6.4)	0 (0)	6 (2.4)
Mar 2019	83 (8.5)	692 (8.3)	271 (11.9)	10,724 (31.4)	595 (10.7)	5930 (5.3)	605 (10.5)	5195 (10.6)	0 (0)	28 (11.4)
April 2019	56 (5.8)	310 (3.7)	165 (7.2)	1097 (3.2)	522 (9.4)	3865 (3.5)	525 (9.1)	3911 (8.0)	2 (11.8)	27 (10.9)
May 2019	116 (11.9)	902 (10.8)	171 (7.5)	1965 (5.7)	565 (10.2)	9929 (8.9)	503 (8.7)	3169 (6.5)	2 (11.8)	18 (7.3)
June 2019	42 (4.3)	424 (5.1)	299 (13.1)	1693 (4.9)	772 (13.9)	4589 (4.1)	686 (11.9)	4861 (9.9)	0 (0)	12 (4.9)
July 2019	40 (4.1)	1965 (23.6)	144 (6.3)	795 (2.3)	428 (7.7)	3782 (3.4)	444 (7.7)	2948 (6.0)	4 (23.5)	25 (10.2)
Aug 2019	176 (18.1)	941 (11.3)	221 (9.7)	3842 (11.2)	562 (10.1)	36,267 (32.6)	388 (6.7)	4635 (9.5)	6 (35.3)	21 (8.5)
Sept 2019	65 (6.7)	333 (3.9)	193 (8.5)	1759 (5.1)	272 (4.9)	6813 (6.1)	452 (7.9)	3580 (7.3)	1 (5.9)	21 (8.5)
Oct 2019	105 (10.8)	599 (7.2)	131 (5.7)	1684 (4.9)	299 (5.4)	5529 (4.9)	390 (6.8)	3324 (6.8)	0 (0)	20 (8.1)
Nov 2019	99 (10.2)	674 (8.1)	280 (12.3)	5405 (15.8)	456 (8.2)	7385 (6.6)	644 (11.2)	8206 (16.8)	2 (11.8)	50 (20.3)
Dec 2019	55 (6.7)	578 (6.9)	105 (4.6)	2318 (6.8)	544 (9.8)	20,672 (18.6)	256 (4.5)	3048 (6.2)	0 (0)	17 (6.9)
Total - 2018, n	748	6095	2217	27,814	4581	50,752	4884	39,959	11	260
Total - 2019, n	971	8342	2282	34,175	5564	111,326	5756	48,830	17	246

[a]HCP: health care provider.

[b]GP: general public.

Impact of Pancreatic Cancer Awareness Month

Pancreatic Cancer Awareness Month did not increase pancreatic cancer mentions by health care providers in any of the 5 domains. However, over the study period of 2 years, mentions by the general public increased for treatment, survivorship, and research. Mentions of the topics of prevention and policy did not increase during Pancreatic Cancer Awareness Month (Figure 3).

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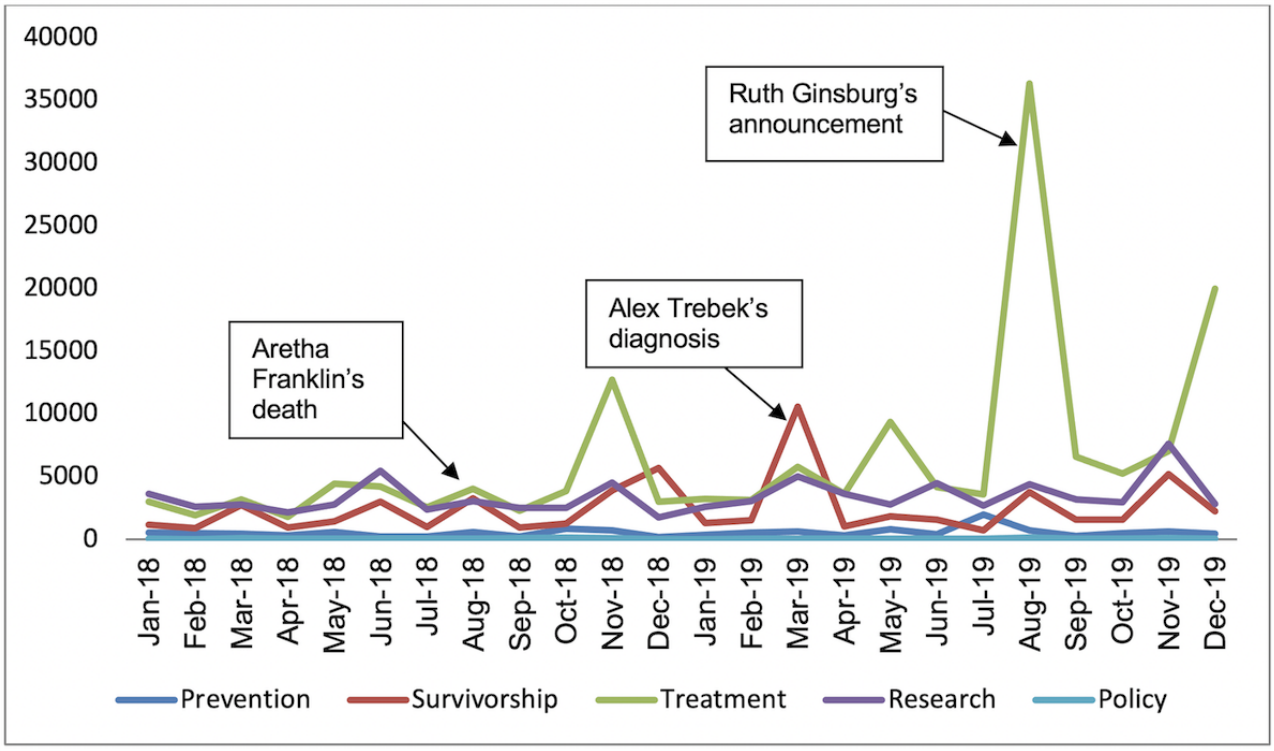
Figure 3. Impact of announcements by public figures of pancreatic cancer diagnoses on Twitter mentions related to pancreatic cancer.



Impact of Announcements by Public Figure of Pancreatic Cancer Diagnoses

We analyzed the impact of announcements by public figures of pancreatic cancer diagnoses on Twitter conversations. Conversations initiated by health care providers did not change with announcements by public figures of pancreatic cancer diagnoses. Among the general public, Mr Trebek's diagnosis was associated with increased conversations about survivorship and Justice Ginsburg's diagnosis was associated with increased conversations about treatment (Figure 3). The announcement of Ms Franklin's death did not result in changes in any of the 5 domains studied as a part of the analysis.

Discussion

Principal Findings

We analyzed Twitter conversations about pancreatic cancer between 2018 and 2019. Twitter discussions by health care providers did not align with discussions initiated by the general public. Pancreatic Cancer Awareness Month did not increase conversations in any of the 5 domains for health care providers, but general public conversations increased in all domains except prevention and policy. Pancreatic cancer announcements by public figures did not affect conversations initiated by health care providers and had varied impact on general public conversations. Mr Trebek's diagnosis increased conversations about survivorship while Justice Ginsberg's announcement increased conversations about treatment.

The current analysis highlights the importance of using social media platforms such as Twitter for analyzing the areas of greatest interest to health care providers and the general public in relation to cancer. The increased interest among the general public in pancreatic cancer treatment could be driven by the low survival rates of patients with pancreatic cancer. Pancreatic cancer is an aggressive malignancy; only about 15% to 20% of patients are diagnosed at an early stage and can benefit from potentially curative resection [11]. Despite advances in recent years, pancreatic cancer treatment continues to remain a formidable challenge. Our findings are in line with other studies that have highlighted the inclination of the general public toward cancer treatment–related discussions on Twitter. A pattern-matched analysis of cancer patients' sentiments on Twitter revealed that patients were most likely to discuss their treatment course (ie, chemotherapy, radiation, and hospital visits). This analysis also identified pancreatic cancer as one of the cancer types associated with the lowest average happiness values among patients [3]. An analysis of Twitter conversations about lung cancer also revealed that users were most likely to tweet about treatment options, which included sharing their personal experiences with treatment or promoting information about newer therapies for lung cancer [12].

The "Twittersphere" also helps in building a communicative and collaborative atmosphere that allows health care providers to involve patients in their care by sharing the latest research and developments in the field [13]. Content experts and researchers can share their work and obtain feedback from the scientific community, patient advocacy groups, and the general public in real time [14]. Live Twitter chats are a unique way for those interested in pancreatic cancer to come together and discuss various topics, including research, policy, and treatment. #PancChat is a Twitter chat that was developed for discussion of relevant information related to pancreatic cancer treatment, diagnosis, and ongoing research with the pancreatic cancer community in a timely manner. #PancChat was developed in 2016 by the Let's Win! Pancreatic Cancer Foundation [15] in

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collaboration with advocacy organizations and a pharmaceutical company. The organizers of the chat develop a series of questions based on the topic being discussed. The event is promoted through various social media platforms and at the time of the chat these questions are serially released. The ensuing conversations can be tracked using the #PancChat hashtag and can be catalogued for future reference. Approximately 20% of the users of #PancChat are patients, advocates, and non–health-care-related individuals. This suggests Twitter can be a powerful tool to disseminate health care information to health care providers, patients, and caregivers [10].

Cancer awareness months are focused on increasing recognition of the disease. Through our analysis, we studied the impact of Pancreatic Cancer Awareness Month on Twitter conversations. We found that Pancreatic Cancer Awareness Month increased conversations initiated by the general public, but that the increase was not uniform from year to year. There was no detectable difference in the domains of prevention and policy. The search algorithm used by our study included both primary prevention and early identification of pancreatic cancer in the prevention domain. There is a growing concern that early detection of pancreatic cancer does not receive adequate attention [16]. A study of Twitter conversations during Breast Cancer Awareness Month found that a majority of the tweets did not prioritize prevention or screening [17]. This suggests that stakeholders should ensure that conversations during Pancreatic Cancer Awareness Month consistently cover various attributes of pancreatic cancer care, including preventative measures. Targeted tweets and conversations specifically related to pancreatic cancer may be essential in increasing discussions on cancer prevention and early identification [10]. The use of machine learning to understand the content and dynamics of conversations related to pancreatic cancer on Twitter will allow the identification of gaps in awareness and communication among health care providers and the general public. This information can then be leveraged to design interventions to address deficiencies and improve communication in those specific areas in a focused manner. This knowledge will also add to the efficiency of targeted interventions such as tailored messaging, which may be used by health care organizations and advocacy groups to further augment dialogue around pancreatic cancer.

Public figure cancer diagnoses have been known to influence public behavior related to cancer. President Ronald Reagan's diagnosis of colon cancer resulted in an increase in the number of colonoscopies performed on asymptomatic individuals [18]. Angelina Jolie's op-ed in the *New York Times* regarding her risk-reducing bilateral mastectomy led to an increase in breast surgery among high-risk women [19]. In the current analysis, we found that a public figure being diagnosed with pancreatic cancer had different impacts on pancreatic cancer–related conversations initiated by the general public, depending on the public figure's personal messaging around the diagnosis and the messaging of reports in the mainstream media. Our findings highlight that public figure diagnoses of pancreatic cancer offer a unique opportunity to capitalize on the increased attention of the general public to the disease. It has also been suggested that public figure cancer announcements can be used to augment conversations about prevention and early diagnosis of cancer [20]. There is a need to study in detail how public figure cancer diagnoses and deaths impact the content and dynamics of Twitter conversations. These data can help physicians, health care systems, and advocacy organizations engage in active communication with targeted audiences and encourage preventative behaviors on a large scale.

Limitations

Limitations of the current study include a short study period and inclusion of tweets or mentions in English only. We did not study regional differences in discussion type. All users not identified as health care providers were identified as the general public, but a more detailed classification of non–health-care providers into patients, survivors, family and friends, advocacy groups, and professional organizations might lead to a better understanding of the conversations initiated by each of these groups. As well, granular details of the conversations could not be harvested or incorporated into the current analysis. Future studies that include a detailed sentiment analysis of the tweets in each domain would allow more insight into the nature and dynamics of Twitter conversations initiated by both health care providers and the general public. Various social media platforms are popular among different groups of users, which means that Twitter users are not representative of the general public. Twitter users are likely to be younger, wealthier, and more educated than the general public [21]. This analysis provides a framework that can be replicated across other social media platforms to gain insight into the conversations taking place about cancer.

Conclusions

This study shows that Twitter conversations initiated by health care providers and the general public are not aligned. Health care providers focus most often on research, while treatment is the most popular topic among the general public. A better understanding of particular areas of interest to the general public might provide researchers, advocacy organizations, and health care systems the opportunity to identify unmet needs related to pancreatic cancer. Pancreatic Cancer Awareness Month increases general public conversations in multiple domains. There is a need to identify and implement strategies to use Pancreatic Cancer Awareness Month to stimulate dialogue that focuses on early detection of pancreatic cancer. Public figure diagnoses or deaths from pancreatic cancer can impact conversations related to pancreatic cancer among the general public. Future studies should also investigate factors that determine how public figure diagnoses impact conversations related to pancreatic cancer.

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Conflicts of Interest

JD is employed by Creation.co. EL reports a research grant from the American Association for Cancer Research (2019 AACR-Novocure Tumor-Treating Fields Research Grant, grant 1-60-62-LOU); past funding from the National Pancreas Foundation and the Minnesota Ovarian Cancer Alliance; an honorarium and travel expenses for a research talk at GlaxoSmithKline (2016); honoraria and travel expenses for lab-based research talks and equipment for laboratory-based research from Novocure (2018-21); an unpaid consultancy for Nomocan Pharmaceuticals; membership on the scientific advisory board, Minnetronix, LLC (2018-present; unpaid); consultant and speaker honoraria, Boston Scientific US (2019); institutional principal investigator for clinical trials, sponsored by Celgene, Novocure, Intima Biosciences, and the NCI; and University of Minnesota membership in the Caris Life Sciences Precision Oncology Alliance (unpaid). AJO has served in a consulting or advisory role for Immunomedics, Inc, Celgene, Tyme Therapeutics, Array, Merck, BMS, ProStrakan, Novartis, Pfizer, Eli Lilly, and Genentech, and has served in the Speaker's Bureau of Daiichi Sankyo. AM receives royalties for a pancreatic cancer biomarker test from Cosmos Wisdom Biotechnology; this financial relationship is managed and monitored by the University of Texas MD Anderson Cancer Center Conflict of Interest Committee. AM is also listed as an inventor on a patent that has been licensed by Johns Hopkins University to Thrive Earlier Detection. AM received a grant from the Sheikh Khalifa bin Zayed Foundation. MSB has performed consulting for AstraZeneca, Merck, Ipsen, Foundation Medicine, Science 37, and Cancer Commons.

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Abbreviations

OR: odds ratio

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Original Paper

Feasibility and Acceptability of a Physical Activity Tracker and Text Messages to Promote Physical Activity During Chemotherapy for Colorectal Cancer: Pilot Randomized Controlled Trial (Smart Pace II)

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Abstract

Background: We conducted a pilot 2-arm randomized controlled trial to assess the feasibility of a digital health intervention to increase moderate-to-vigorous physical activity in patients with colorectal cancer (CRC) during chemotherapy.

Objective: This study aimed to determine whether a digital health physical activity intervention is feasible and acceptable during chemotherapy for CRC.

Methods: Potentially eligible patients with CRC expected to receive at least 12 weeks of chemotherapy were identified in person at the University of California, San Francisco, and on the web through advertising. Eligible patients were randomized 1:1 to a 12-week intervention (Fitbit Flex, automated SMS text messages) versus usual care. At 0 and 12 weeks, patients wore an Actigraph GT3X+ accelerometer for 7 days and completed surveys, body size measurements, and an optional 6-minute walk test. Participants could not be masked to their intervention arm, but people assessing the body size and 6-minute walk test outcomes were masked. The primary outcomes were adherence (eg, Fitbit wear and text response rate) and self-assessed acceptability of the intervention. The intervention would be considered feasible if we observed at least 80% complete follow-up and 70% adherence and satisfaction, a priori.

Results: From 2018 to 2020, we screened 240 patients; 53.3% (128/240) of patients were ineligible and 26.7% (64/240) declined to participate. A total of 44 patients (44/240, 18%) were randomized to the intervention (n=22) or control (n=22) groups. Of these, 57% (25/44) were women; 68% (30/44) identified as White and 25% (11/44) identified as Asian American or Pacific Islander; and 77% (34/44) had a 4-year college degree. The median age at enrollment was 54 years (IQR 45-62 years). Follow-up at 12 weeks was 91% (40/44) complete. In the intervention arm, patients wore Fitbit devices on a median of 67 out of 84 (80%) study days and responded to a median of 17 out of 27 (63%) questions sent via SMS text message. Among 19 out of 22 (86%) intervention patients who completed the feedback survey, 89% (17/19) were satisfied with the Fitbit device; 63% (12/19) were satisfied with

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the SMS text messages; 68% (13/19) said the SMS text messages motivated them to exercise; 74% (14/19) said the frequency of SMS text messages (1-3 days) was ideal; and 79% (15/19) said that receiving SMS text messages in the morning and evening was ideal.

Conclusions: This pilot study demonstrated that many people receiving chemotherapy for CRC are interested in participating in digital health physical activity interventions. Fitbit adherence was high; however, participants indicated a desire for more tailored SMS text message content. Studies with more socioeconomically diverse patients with CRC are required.

Trial Registration: ClinicalTrials.gov NCT03524716; https://clinicaltrials.gov/ct2/show/NCT03524716

(JMIR Cancer 2022;8(1):e31576) doi:10.2196/31576

KEYWORDS

exercise; treatment; colon cancer; rectal cancer; digital health; wearables; SMS

Introduction

Background

Colorectal cancer (CRC) is the fourth most diagnosed cancer and the second leading cause of cancer-related deaths in the United States [1]. Prospective studies suggest that physical activity after CRC diagnosis is associated with longer survival, including in patients with advanced or metastatic disease [2-5]. Moreover, patients with nonmetastatic CRC who engage in less physical activity after diagnosis have a 32% increased risk of CRC-specific mortality compared with patients who maintain their prediagnosis levels of activity [6]. Given that physical activity tends to decline during treatment [7], interventions that help patients with CRC to maintain their physical activity levels during treatment may be important adjuncts to standard oncological therapies.

Several interventions are being evaluated for their impact on physical activity in patients with CRC [8]. The Colon Health and Life-Long Exercise Change (CHALLENGE) and Focus on Reducing Dose-limiting Toxicities in Colon Cancer with Resistance Exercise (FORCE) trials are 2 examples of such interventions [8,9]. CHALLENGE is an active randomized controlled trial examining the effects of a structured exercise program on disease-free survival among patients with high-risk stage 2 or 3 colon cancer who have completed adjuvant chemotherapy [8]. FORCE is an open randomized controlled trial examining the effects of resistance training on relative dose intensity and chemotoxicities in patients with nonmetastatic colon cancer receiving adjuvant chemotherapy [9]. Notably, CHALLENGE was designed as a supervised program, and FORCE focused on resistance training; both studies enrolled only patients with nonmetastatic colon cancer. Indeed, most studies to date have focused on people with nonmetastatic disease and those who have already completed treatment. There remains a need to determine the feasibility of physical activity interventions for patients with CRC during active treatment. Moreover, participation in supervised exercise intervention programs for patients with cancer may be limited by time, expense, and access to treatment centers offering exercise services. Thus, remotely delivered interventions may increase the accessibility of exercise interventions for patients with CRC.

Previous Work

Digital health tools, such as physical activity trackers, SMS text messaging, and apps, offer low-cost approaches to increase physical activity [10]. One study evaluated adherence to wearing a Fitbit in patients with early breast cancer on chemotherapy and concluded that additional intervention components, such as phone calls, SMS text messages, or other reminders, are needed to maintain adherence to wearing the Fitbit [11]. Few studies have evaluated similar intervention components in patients with CRC, especially those undergoing chemotherapy. A review of consumer wearable health intervention studies with survivors of breast cancer, prostate cancer, and CRC identified 8 randomized controlled trials conducted among people with these cancers; only one of these trials (Smart Pace I), conducted by our team, focused exclusively on survivors of CRC [12]. In that study, we reported that digital health tools, including a Fitbit Flex and SMS text messages, were feasible, were acceptable, and may increase physical activity among survivors of CRC after completion of chemotherapy [13].

Objectives

In this study (Smart Pace II), we aim to determine whether a digital health physical activity intervention is feasible and acceptable during chemotherapy, with the goal to prevent the decline in physical activity that often occurs during treatment for CRC. We conducted a 12-week pilot 2-arm randomized controlled trial with patients with CRC receiving chemotherapy. Our primary objective is to evaluate the feasibility and acceptability of a digital physical activity intervention in this patient population. In addition, we sought to estimate the effect of the intervention on physical activity, cardiorespiratory fitness estimated through the 6-minute walk test distance, body weight, and blood pressure from enrollment to 12 weeks.

Methods

Smart Pace II was a 2-arm (1:1) pilot randomized controlled trial. The study was approved by the institutional review board of the University of California, San Francisco (UCSF).

Study Population and Recruitment

Overview

We recruited individuals with colon or rectal cancer who were recommended to receive at least 12 weeks of chemotherapy. Potentially eligible participants were identified through the Gastrointestinal Oncology Clinic at UCSF as well as through public advertising on the web, at community events, and in local oncology clinics. Potential participants at the UCSF were approached in person and by email. The intervention was

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administered remotely, and recruitment was not restricted to individuals receiving chemotherapy at the UCSF. Eligibility criteria included the expectation of receiving at least 12 weeks of chemotherapy, the ability to speak and read English, access to a mobile phone with email and SMS text messaging capabilities, ≥4 weeks since the last major surgery, and provider endorsement of patient safety to participate in unsupervised moderate physical activity. Patients were excluded if they self-reported ≥150 minutes per week of moderate-to-vigorous physical activity (MVPA) on the modified Godin Leisure Time Exercise Questionnaire or had contraindications to exercise at the time of enrollment [14]. We initially excluded participants who owned a physical activity tracker designed to be worn all day (not just during exercise sessions), such as a Fitbit. In June 2019, we refined this criterion to exclude people who owned physical activity trackers and had worn them in the past month; people who owned trackers but were not wearing them would still be eligible. The eligibility criterion that excluded people who owned and wore a physical activity tracker was completely removed in August 2019.

Between March 1, 2018, and March 17, 2020, a total of 240 patients were assessed for eligibility (Figure 1). Of these 240 people, 26.7% (64/240) declined to participate. Interested patients were asked to complete a web-based screening survey using Research Electronic Data Capture (REDCap) [15]. We then contacted the treating provider for each potential participant to confirm clinical eligibility and endorsement of the patient's safety to engage in unsupervised moderate physical activity. One provider did not respond, so we were unable to ascertain eligibility for one potential participant. Following these screening procedures, 53.8% (128/240) of the patients were deemed ineligible. The main reasons were lack of provider approval (64/128, 50%), a treatment plan that did not match the eligibility requirements (24/128, 18.8%), medical contraindications to exercise (16/128, 12.5%), or self-reported exercise of ≥150 minutes per week of MVPA (24/128, 18.8%). One patient passed away during the screening period and one patient was not allowed to wear the Fitbit at work. Thus, after recruitment and screening, 18.8% (45/240) of screened patients were considered eligible for participation.

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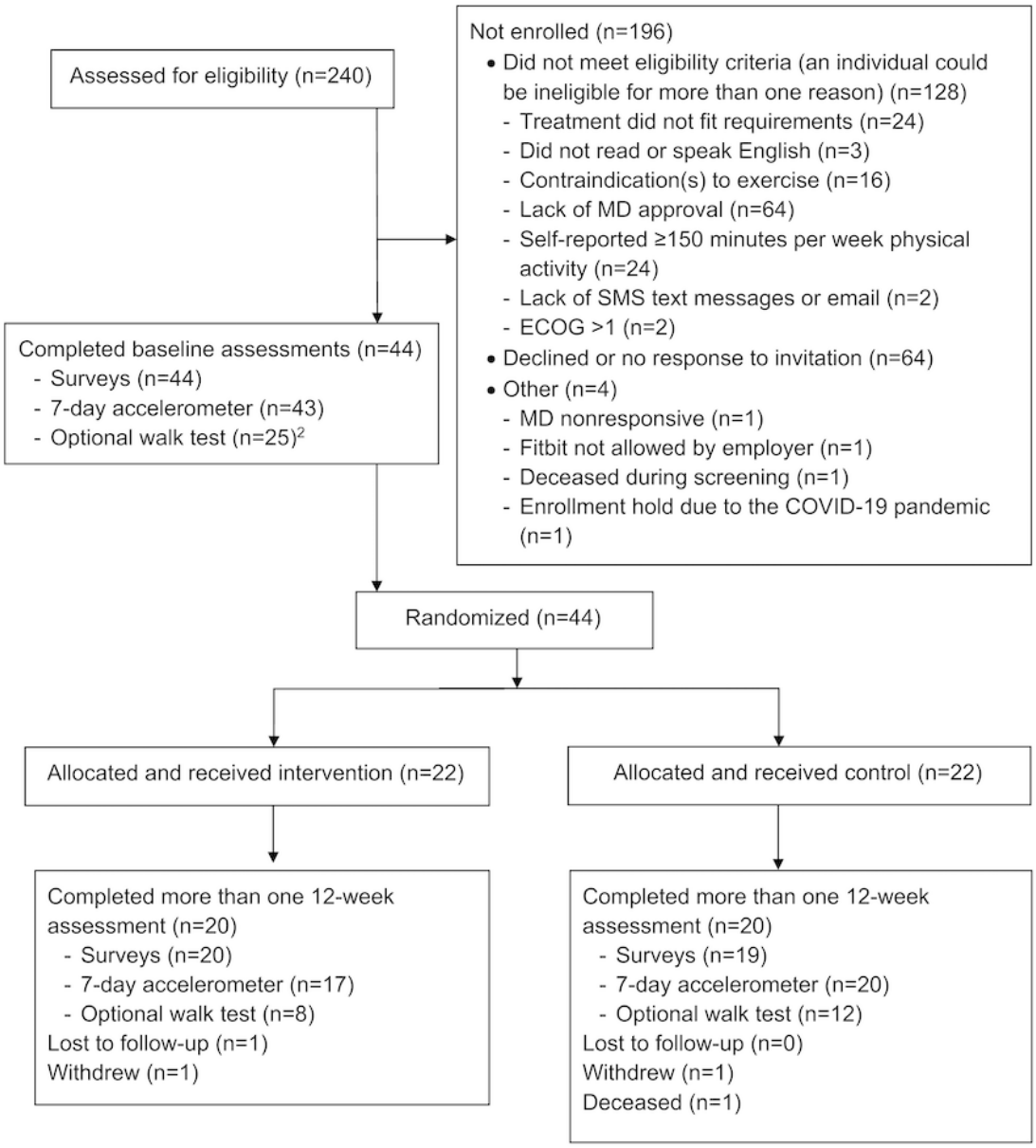
Figure 1. CONSORT (Consolidated Standards of Reporting Trials) flow diagram for the Smart Pace II study, a randomized controlled pilot study evaluating a 12-week physical activity intervention for people receiving chemotherapy for colon or rectal cancer. Stay Home Public Orders were enacted on March 17, 2020, in San Francisco, California, and all elective medical visits were cancelled, including two baseline and five 12-week 6-minute walk tests. ECOG: Eastern Cooperative Oncology Group; MD: medical doctor.



Consent and Randomization

Once participants were confirmed as eligible, informed consent was obtained either in person or electronically using DocuSign. Between March 15, 2018, and March 20, 2020, a total of 44 participants were randomized 1:1 to intervention or control, using a computer-generated randomization scheme created by a blinded study statistician (LZ). The 45th interested and eligible participant was not randomized owing to an enrollment hold as a result of the COVID-19 pandemic. The randomization scheme was uploaded to REDCap, and the study research coordinator used REDCap to determine a given participant's assigned intervention arm. Relevant study materials were then distributed to the participants in person or by mail by the study research coordinator.

Interventions

Intervention Arm

Participants in the intervention arm received a printed booklet about physical activity after cancer, daily fully automated SMS text messages (see Multimedia Appendix 1 for sample SMS text messages), a Fitbit Flex 2 Fitness Wristband (hereafter referred to as the Fitbit), and a list of home-based exercise apps and videos. The intervention was intended to be stand-alone with no human involvement. Participants received written instructions on how to set up the Fitbit and were asked to wear their Fitbit on their wrist every day during the 12-week study period; they were allowed to keep the Fitbit after the study. To receive the SMS text messages automatically during the study, participants' phone numbers were registered by a research coordinator on a custom-built Drupal website that interacted

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with Twilio to facilitate sending and receiving SMS text messages. Participants were encouraged to work up to the United States Physical Activity Guidelines of 150 minutes per week of MVPA through the SMS text messages [16]. A total of 21 SMS text messages specifically promoted aerobic exercise, 10 specifically mentioned resistance exercise, and 2 SMS text messages specifically encouraged flexibility exercise. Notably, 4 SMS text messages asked participants, "Good Morning! How is your energy level today? Text back 'H' if you feel great, 'M' if you feel ok, and 'L' if you feel very tired." Tailored feedback for the day's activity was sent based on the participants' responses. For example, if the participant replied "L," they received the following message: "(1/2) You are going through a lot. Sometimes light exercise can help you feel better. (2/2) Walking or yoga are good options—try to do just 10 minutes today at an easy and comfortable pace and see if that helps!" A total of 6 SMS text messages prompted the participants to wear and synchronize their Fitbit devices. Owing to the nature of the intervention, the participants were not blinded to their assigned intervention arm.

Control Arm

Participants in the control arm received a printed booklet about physical activity for cancer survivors after randomization and were given a Fitbit after completion of the 12-week follow-up assessments to compensate for study participation.

Study Measures

Feasibility

We assessed the feasibility of the intervention by calculating the median number of days that intervention participants wore the Fitbit; the median number of SMS text messages that asked for a reply that intervention participants responded to; and the proportion of the study participants who completed at least one 12-week follow-up survey, overall and by arm. We counted the Fitbit as worn on a given day if >1500 steps were recorded [17]. SMS text message adherence was calculated as the mean proportion of texts that requested a reply to which each intervention participant responded. We stated that we would consider the intervention to be feasible if we achieved at least 70% adherence on average (Fitbit worn at least 59 days out of the 84 study days; 19 or more text messages responded to out of 27 that asked for a reply) and if 80% of participants completed at least one 12-week follow-up survey, a priori.

Acceptability

The acceptability of the intervention was evaluated by an investigator-created questionnaire administered at 12 weeks on the web using REDCap [15]. Intervention participants were asked to what degree they agreed with statements regarding the intervention components (eg, SMS text messages and Fitbit). Responses were coded on a 5-point Likert scale (eg, 1=strongly agree, 2=agree, 3=undecided, 4=disagree, and 5=strongly disagree). The questionnaire also included 2 open-ended questions for other feedback on the SMS text messages and Fitbit devices.

Physical Activity

Participants' physical activity was assessed as a secondary outcome. Activity was measured using ActiGraph GTX3+ accelerometers (ActiGraph LLC) worn on the wrist for 7 consecutive days at enrollment and 12 weeks [18]. Data were recorded and analyzed in 5-second epochs. A minimum of 3 days with a valid wear time of at least 10 hours was required for inclusion in the analysis [19,20]. To determine valid hours, nonwear time was identified using the Troiano 2007 algorithm in the ActiLife software (version 6.13.4).

After the study was completed, we used the Freedson Adult 1998 cutoff points to identify the average minutes per day of sedentary (0-100 counts per minute), light (101-1952 counts per minute), moderate (1953-5724 counts per minute), hard (5725-9498 counts per minute), and very hard (9499-16,000 counts per minute) physical activity [21]. We also estimated minutes per week spent in at least 10-minute bouts of MVPA. To do so, we divided the total time in Freedson Adult 1998 bouts calculated by the ActiLife software by the number of calendar days with valid wear time and multiplied by 7. These calculations were performed after the study was completed, so participants and researchers were blinded to the baseline accelerometer-assessed physical activity minutes per week values at the time of randomization.

6-Minute Walk Test, Body Weight, and Blood Pressure

At enrollment and 12 weeks, participants who were able to come to the UCSF were given the option to complete a 6-minute walk test, a submaximal test correlated with peak VO_2 and widely used to detect changes in exercise tolerance in adults [22]. If the test was performed on the same day as the scheduled treatment, the 6-minute walk test was performed before the administration of chemotherapy. Data on participants' body weight and blood pressure were abstracted from participants' medical records (patients from UCSF) or obtained from participants' providers (patients not from UCSF) at baseline and 12 weeks.

Adverse Events

A survey was created by the investigator team to collect self-reported adverse events during the intervention period. Participants completed a brief *health check-in* on the web at 0, 4, 8, and 12 weeks using REDCap surveys delivered via email. The survey queried recent chemotherapy treatments, current body weight, medication use, hospitalizations, and whether the patient had experienced any of the following conditions in the past 4 weeks: low back pain, knee pain, shoulder pain, arthritis, chest pain, shortness of breath, fatigue, leg cramping, muscle pain, and dizziness or vertigo. If participants reported any of these conditions, they were asked to report the onset and duration of symptoms, whether any activities made it better or worse, and if they took any medication for the condition.

Sample Size

Our target sample size of 48 participants was based on the number of participants in previous pilot studies [13]. This number was sufficient to answer our primary objective of feasibility, quantified using Fitbit adherence (number of days

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that the participants wore the device) and text message response (number of replies to SMS text messages that asked for a reply). We stopped the trial in March 2020, after 44 participants were randomized, owing to the COVID-19 pandemic.

Statistical Analysis

Descriptive statistics, including counts, percentages, means, SDs, medians, and ranges were used to describe participant characteristics and reports of adverse events. All statistical analyses were conducted using R [23].

We conducted 1-sample *Z* tests to determine whether the observed adherence was significantly less than the a priori cutoff of 70%. We also used 1-sample *Z* tests to determine whether the proportion of the study participants (overall and by group) that completed a 12-week follow-up survey was significantly less than a prior cutoff of 80% or more. Fisher exact test was used to compare attrition between the 2 arms. We reported the participants' responses to the feedback questionnaire using descriptive statistics.

The secondary effects of the intervention from baseline to 12 weeks within and between the intervention and control arms were estimated using weighted *t* tests for physical activity measures and Mann–Whitney tests for body weight, blood pressure, and the 6-minute walk test.

Results

We randomized 44 participants with CRC to the intervention (n=22) or control (n=22) arms (Figure 1) between March 2018 and March 2020. The assigned intervention was administered to all 44 participants. Follow-up at 12 weeks was 91% (20/22) complete in both arms. In the intervention arm, one participant withdrew, reporting that the study was incompatible with the chemotherapy schedule and citing the inconvenience of charging and syncing the Fitbit. One intervention arm patient was lost to follow-up for unknown reasons. In the control arm, 1 participant died during the intervention phase because of cancer progression, and 1 participant withdrew after transferring care to another treatment facility.

Study Population Characteristics

The characteristics of the intervention and control arms are listed in Table 1. Most participants (29/44, 66%) were enrolled at the start of their first line of chemotherapy, and 6 were receiving their third or more line of chemotherapy (6/44, 14%). The individuals enrolled with an initial diagnosis of stage 1 or 2 disease were receiving neoadjuvant chemotherapy (1 person), adjuvant chemotherapy to reduce the risk of recurrence (3 people), or chemotherapy for recurrent disease (1 person). The intervention and control groups had a similar median age at enrollment and similar gender and cancer site, stage, and treatment distributions. However, by chance owing to the small sample size, a higher proportion of the control group were patients from UCSF who identified as Asian American or Pacific Islander; the median BMI of this group was also lower than that of the intervention group.

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Table 1. Demographic characteristics and clinical factors of participants with colorectal cancer undergoing chemotherapy in a 2-arm pilot randomized controlled trial of a 12-week digital physical activity intervention (N=44).

Characteristics	Total	Intervention	Control
Participants, n (%)	44 (100)	22 (50)	22 (50)
Patients from UCSF[a], n (%)	37 (84)	15 (34)	22 (50)
Age at enrollment (years), median (IQR)	54 (45-62)	53 (41-59)	53 (47-67)
Females, n (%)	25 (57)	14 (32)	11 (25)
BMI (kg/m^2), median (IQR)	25.7 (21.5-28.7)	27.5 (22.7-30.5)	24.0 (20.8-26.9)
Highest level of education, n (%)			
2-year college or less	10 (23)	6 (14)	4 (9)
4-year college	16 (36)	9 (20)	7 (16)
Graduate or professional degree	18 (41)	7 (16)	11 (25)
Self-identified race or origin[b], n (%)			
Asian American or Pacific Islander	11 (25)	3 (7)	8 (18)
White	30 (68)	18 (41)	12 (27)
Other or unknown	4 (9)	2 (5)	2 (5)
Primary cancer site, n (%)			
Colon	28 (64)	12 (27)	16 (36)
Rectum	16 (36)	10 (23)	6 (14)
Months since diagnosis, median (IQR)	4 (2-19)	4 (2-6)	4 (2-8)
Stage at diagnosis, n (%)			
1-2	5 (11)	3 (7)	2 (5)
3	22 (50)	10 (23)	12 (27)
4	17 (39)	9 (21)	8 (18)
Treatments received for colon or rectal cancer at the time of enrollment (all that apply), n (%)			
Surgery	28 (64)	14 (32)	14 (32)
Radiation	5 (11)	3 (7)	2 (5)
Systemic chemotherapy	44 (100)	22 (50)	22 (50)
Other	1 (2)	0	1 (2)
Ostomy status at enrollment, n (%)			
No ostomy	32 (73)	16 (36)	16 (36)
Permanent ostomy	6 (14)	4 (9)	2 (5)
Previously reversed ostomy	2 (5)	1 (2)	1 (2)
Ostomy awaiting reversal	4 (9)	1 (2)	3 (7)
Current line of chemotherapy, n (%)			
1	29 (66)	13 (30)	16 (36)
2	9 (20)	4 (9)	5 (11)
≥3	6 (14)	5 (11)	1 (2)
Disease status at enrollment, n (%)			
No evidence of disease	5 (11)	1 (2)	4 (9)
Stable disease	18 (41)	10 (23)	8 (18)
Progressive disease	21 (48)	11 (25)	10 (23)
Smoking status, n (%)			

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Characteristics	Total	Intervention	Control
Never	30 (68)	13 (30)	17 (39)
Former	14 (32)	9 (20)	5 (11)
Comorbidities[b], median (IQR)	1 (0-2)	1 (1-1)	1 (1-1)
Comorbid conditions[b], n (%)			
High blood pressure	13 (30)	5 (11)	8 (18)
Elevated cholesterol	13 (30)	6 (14)	7 (16)
Cancer (not including CRC[c])	7 (16)	5 (11)	2 (5)
Arthritis	5 (11)	3 (7)	2 (5)
Diabetes mellitus	4 (9)	1 (2)	3 (7)
Venous thromboembolism	4 (9)	2 (5)	2 (5)
Chronic kidney disease	3 (7)	1 (2)	2 (5)
Asthma	2 (5)	1 (2)	1 (2)
Other comorbid conditions[d]	6 (14)	2 (5)	4 (9)

[a]UCSF: University of California, San Francisco.[b]Comorbid conditions were ascertained using self-report.

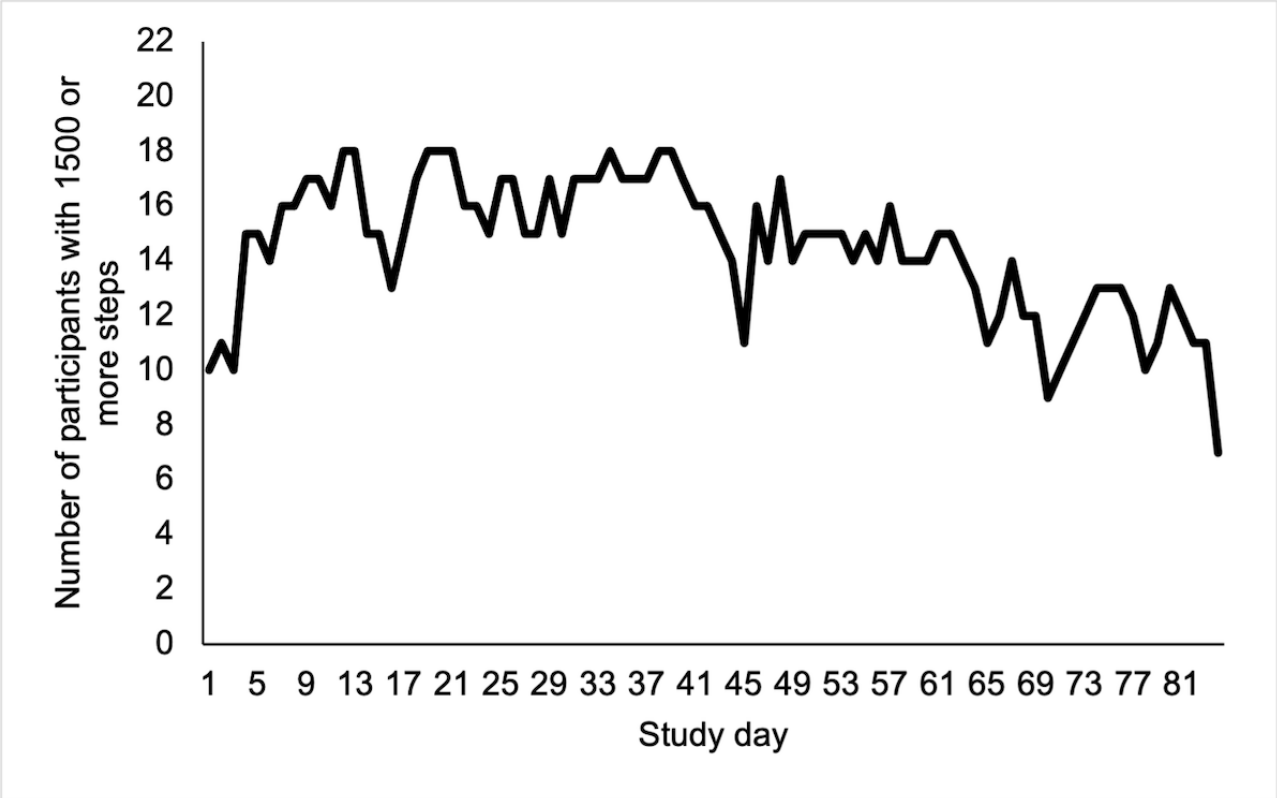
[c]CRC: colorectal cancer.

[d]Other comorbidities reported by 1 person each included transient ischemic attack, stroke, osteoporosis, history of hip fracture, multiple sclerosis, emphysema, or chronic bronchitis.

Adherence and Attrition

Participants randomized to the intervention arm wore their Fitbits for a median of 67 out of 84 study days (IQR 53-80 days). A total of 2 participants never wore the Fitbit, and 2 participants had <10 days of wear time. A total of 6 participants had >80 days of wear time. Fitbit use trended down slightly over time (Figure 2). There was no correlation between age and gender of the participants and wear time. Participants with stage 4 cancer had a median Fitbit wear time of 56 days (IQR 47-76 days) compared with a median of 77 days among participants with stage 1 to 3 disease (IQR 56-82).

Figure 2. Number of participants in the intervention arm of the Smart Pace II pilot study who recorded at least 1500 steps per day on the Fitbit, by study day (n=22).



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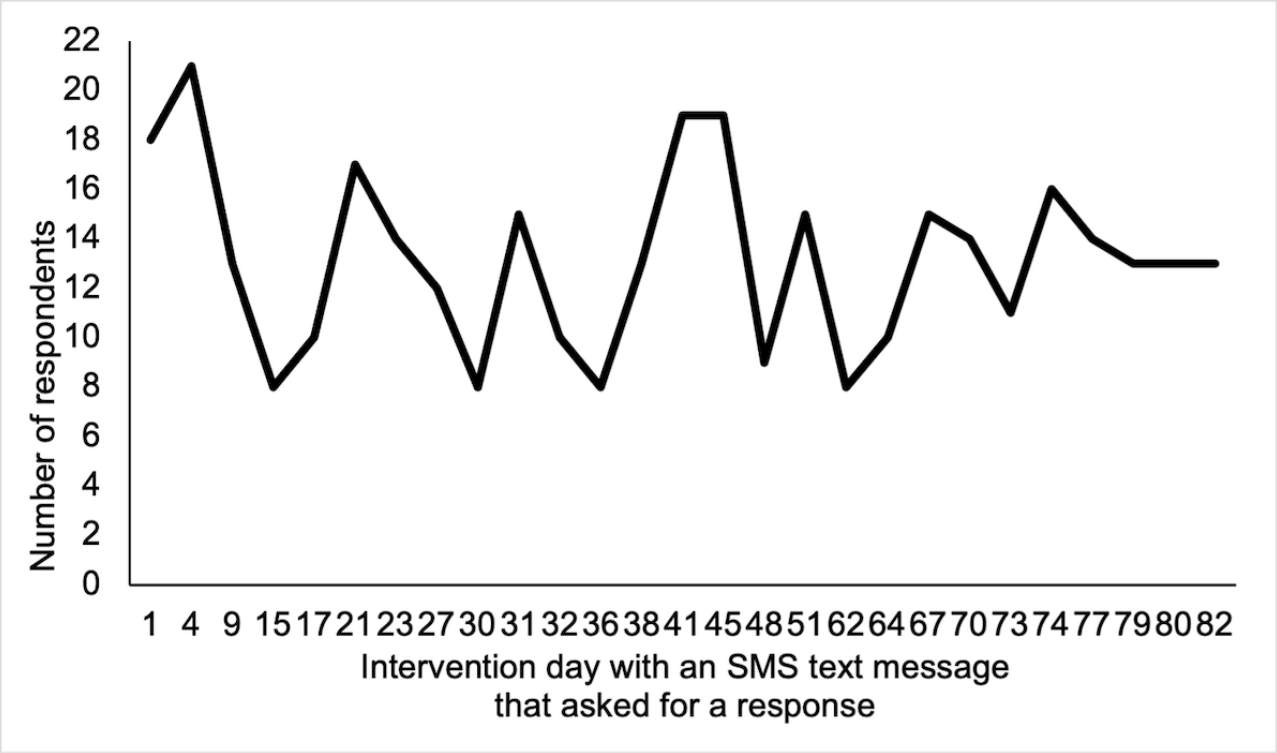
Overall, participants in the intervention arm responded to a median of 17 out of 27 SMS text messages that asked for a reply (63%; IQR 12-23; range 1-26). SMS text message response rates fluctuated over time (Figure 3). SMS text messages sent on days 15, 36, and 62, which queried whether participants had achieved the goals they were asked to set at the beginning of the study on day 8, were among the messages with the lowest response rates. No patterns were observed regarding the content of SMS text messages that received the highest response rates. SMS text message response rates did not vary by age, gender, or cancer stage.

Figure 3. Number of participants in the intervention arm who responded to the SMS text messages that asked for a reply in the Smart Pace II pilot study (n=22).



Acceptability

Most participants reported that the intervention was acceptable (Table 2 and Table 3). Out of the 22 participants in the intervention arm, 19 (86%) completed the feedback questionnaire. Among the respondents, 63% (12/19) reported satisfaction with the SMS text messages overall and 89% (17/19) reported satisfaction with the Fitbit and an expectation that they would continue to wear the Fitbit after the study ended.

Table 2. Overall satisfaction with 12 weeks of SMS text messages and a Fitbit Flex 2 among individuals receiving chemotherapy for colorectal cancer (n=22).

Responses	Very satisfied	Satisfied	Neutral	Dissatisfied	Very dissatisfied	Missing
Overall satisfaction with text messages, n (%)	4 (18)	8 (36)	6 (27)	1 (5)	0	3 (14)
Overall satisfaction with Fitbit, n (%)	7 (32)	10 (46)	1 (5)	1 (5)	0	3 (14)

When asked about specific features (Table 3), 68% (13/19) agreed that the SMS text messages motivated them to exercise and that the content was interesting; 74% (14/19) said that the frequency of the messages was ideal (1 every 1-3 days), and 79% (15/19) said that the timing of the messages was ideal (morning and evening). The most frequent recommendation for improvement was to improve the personalization of messages (Multimedia Appendix 2). Regarding the Fitbit, 2 participants said that they did not like using a wearable device or did not feel the need to track their activities daily. Additional feedback from participants included difficulty adhering to the intervention because of treatment-related fatigue and restrictions imposed during the COVID-19 pandemic.

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Table 3. Responses to the feedback survey regarding acceptability of 12 weeks of SMS text messages and a Fitbit Flex 2 among individuals receiving chemotherapy for colorectal cancer (n=22 participants).

Responses	Strongly agree	Agree	Undecided	Disagree	Strongly disagree	Missing
Text messages motivated me to exercise, n (%)	3 (14)	10 (46)	1 (5)	4 (18)	1 (5)	3 (14)
Content of text messages was interesting, n (%)	3 (14)	10 (46)	3 (14)	1 (5)	2 (9)	3 (14)
Frequency of text messages was ideal, n (%)	6 (27)	8 (36)	2 (9)	1 (5)	2 (9)	3 (14)
Timing of text messages was ideal, n (%)	3 (14)	12 (55)	2 (9)	1 (5)	1 (5)	3 (14)
Fitbit motivated me to exercise, n (%)	7 (32)	9 (41)	1 (5)	2 (9)	0 (0)	3 (14)

Estimated Changes in Physical Activity

Physical activity levels measured by the accelerometer at enrollment and 12 weeks for participants in the intervention and control groups are shown in Multimedia Appendix 3. No patient recorded any vigorous physical activity at any time point. Overall, on average, the participants accumulated 110 minutes per week (SD 103 minutes per week) of moderate-intensity activity in bouts of 10 minutes or longer at enrollment. By chance, the intervention arm recorded more time in moderate activity bouts compared with controls at enrollment (mean 141.5, SD 115.5 minutes per week and mean 80.7, SD 83.5 minutes per week, respectively).

When examining changes from 0 to 12 weeks, both the intervention and control groups decreased their physical activity on average over the 12-week study period. The intervention arm had a mean reduction in moderate activity accumulated in 10-minute bouts of 21.3 minutes per week (SD 144.8 minutes per week); the control arm had a mean reduction in moderate activity accumulated in 10-minute bouts of 16.3 minutes per week (SD 121.2 minutes per week). There was no difference in the change in moderate activity accumulated in bouts of 10 minutes when comparing the 2 groups (mean difference 0.2, SD 6.2 minutes per week). When examining individual changes, 47% (8/17) of participants in the intervention arm and 35% (7/20) in the control arm with data at both time points increased bouts of moderate activity from enrollment to 12 weeks by at least 1 minute. Notably, when examining total activity throughout the day (not specifically ≥10-minute bouts), participants in the intervention arm reduced moderate activity, light activity, and steps more than the control arm. Finally, the total time moving at a moderate intensity was high in both the intervention and control arms and the change in activity between time points was highly variable with wide SDs.

Estimated Changes in 6-Minute Walk Test, Body Weight, and Blood Pressure

The 6-minute walk test, body weight, and blood pressure at enrollment and 12 weeks for participants in the intervention and control groups are shown in Multimedia Appendix 3. Participants in both arms increased their 6-minute walk test distance by an average of 37 meters (SD 39 meters) in the intervention group and 46 meters (SD 59 meters) in the control group. For body weight, the intervention group had a mean change of −0.8 pounds (SD 5.7 pounds), whereas the control group had a mean change of 0.1 pounds (SD 9.5 pounds). When examining individual changes, we observed that 65% (11/17) participants in the intervention arm with data available at both time points lost weight from enrollment to 12 weeks, whereas 42% (8/19) participants in the control arm lost weight from 0 to 12 weeks. There were no significant changes in blood pressure within or between the 2 groups. The average difference in systolic blood pressure from 0 to 12 weeks for the intervention group was 6.4 mm Hg (SD 12.2 mm Hg); and the control group had a mean change of −0.4 mm Hg (SD 10.6 mm Hg). The average difference in diastolic blood pressure for the intervention group was −0.5 mm Hg (SD 9.6 mm Hg); the control group had a mean change of −5.8 mm Hg (SD 9.0 mmm Hg). When examining individual changes, 53% (9/17) participants in the intervention arm and 67% (12/18) participants in the control arm decreased their systolic and diastolic blood pressure from 0 to 12 weeks. As with the physical activity data, there was considerable variability in responses between participants.

Adverse Events

The number of reported adverse events is presented in Table 4. There were no serious adverse events related to the intervention, and the intervention did not appear to increase reports of nonserious adverse events compared with baseline. A total of 4 participants in the control group reported hospitalizations during the study, and 1 participant in the control group passed away during the study because of cancer progression. There were no hospitalizations or deaths in the intervention group.

For nonserious adverse events, fatigue was the most reported adverse event during the study, but the number of times fatigue was reported was highest at enrollment and it did not increase during the intervention period. In addition, as described above, 4 of the SMS text messages in the intervention arm asked participants to rate how they felt (days 4, 31, 45, and 67). On day 4, 32% (7/22) of the participants in the intervention arm responded saying they were very tired, 27% (4/15) said they were very tired on day 31, 11% (2/19) said they were very tired on day 45, and 27% (4/15) said they were very tired on day 67.

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Table 4. Adverse events reported among participants receiving chemotherapy and participating in a 12-week digital physical activity intervention[a] (N=44).

Adverse events	Intervention, n (%)				Control, n (%)			
	Before enrollment (n=22)	0-4 weeks (n=17)	5-8 weeks (n=20)	9-12 weeks (n=19)	Before enrollment (n=22)	0-4 weeks (n=17)	5-8 weeks (n=15)	9-12 weeks (n=19)
Total adverse events	51	40	48	38	38	31	20	22
Low back pain	7 (14)	4 (10)	5 (10)	6 (16)	4 (11)	1 (3)	1 (5)	2 (9)
Knee pain	2 (4)	2 (5)	2 (4)	1 (3)	1 (3)	0 (0)	0 (0)	1 (5)
Shoulder pain	3 (6)	2 (5)	5 (10)	2 (5)	4 (11)	1 (3)	0 (0)	0 (0)
Inflammation of the joints	2 (4)	2 (5)	2 (4)	0 (0)	1 (3)	2 (6)	1 (5)	1 (5)
Chest pain	0 (0)	1 (3)	2 (4)	1 (3)	1 (3)	0 (0)	0 (0)	0 (0)
Shortness of breath	5 (10)	3 (8)	4 (8)	5 (13)	2 (5)	0 (0)	0 (0)	1 (5)
Fatigue	18 (35)	13 (33)	16 (33)	14 (37)	12 (32)	13 (42)	11 (55)	12 (55)
Leg cramping	4 (8)	3 (8)	2 (4)	1 (3)	2 (5)	4 (13)	1 (5)	1 (5)
Muscle pain	4 (8)	3 (8)	4 (8)	2 (5)	2 (5)	3 (10)	1 (5)	0 (0)
Dizziness or vertigo	2 (4)	1 (3)	3 (6)	2 (5)	2 (5)	2 (6)	1 (5)	1 (5)
Other orthopedic limitation	1 (2)	2 (5)	0 (0)	0 (0)	1 (3)	2 (6)	2 (10)	1 (5)
Doctor's visit, excluding standard cancer follow-up	3 (6)	4 (10)	3 (6)	4 (11)	3 (8)	2 (6)	2 (10)	1 (5)
Hospitalization[b]	0 (0)	0 (0)	0 (0)	0 (0)	3 (8)	1 (3)	0 (0)	0 (0)
Death[c]	N/A[d]	0 (0)	0 (0)	0 (0)	N/A	0 (0)	0 (0)	1 (5)

[a]Participants were asked at the time of enrollment to report if they had experienced any adverse events in the past month. The survey was repeated at 4, 8, and 12 weeks.

[b]Reasons for hospitalization in the month before enrollment included anemia, infection, and fever after receipt of chemotherapy, and at 0-4 weeks, stomach perforation.

[c]One participant in the control arm expired while enrolled in the study because of cancer progression.

[d]N/A: not applicable.

Discussion

Principal Findings

Overall, we observed that a remotely delivered physical activity intervention that included a wristband for self-monitoring physical activity and SMS text messages during chemotherapy for CRC was feasible and acceptable. Although this study was not powered to detect changes in physical activity, our pilot data show a nonstatistically significant decrease in moderate activity accumulated in bouts of at least 10 minutes in both arms (16-21 minutes per week).

Comparison With Previous Work

Notably, the findings from this study with participants who were actively receiving chemotherapy differed from our previous study in people who had previously completed treatment for CRC. In our previous study (Smart Pace I), we observed an average increase in physical activity in participants in the intervention arm [13]. The main difference in our SMS text message content for this study (Smart Pace II) was the addition of questions about how participants felt and tailored activity advice in response. This modification was based on our expectation that participants would feel fatigued during chemotherapy and need support or motivation to promote activity. Messaging to *take it easy* or *build up slowly* sent to a group of people who were active at baseline and felt tired on treatment may have unintentionally contributed to why the intervention arm decreased activity levels slightly more than the control arm, which did not receive SMS text messaging. Further research is required to evaluate whether such messages would have the intended beneficial effect in a sedentary population (encouraging those who feel tired to do a light activity vs nothing). In addition, delivering more nuanced messages that encourage active people to stay active during treatment even when they are tired, without pushing them too far, is a challenge for automated intervention approaches such as SMS text messaging.

Few other studies have conducted remote physical activity interventions in patients with CRC or survivors. Kim et al [24] reported that a home-based exercise intervention with weekly supervised components (counseling or training sessions) significantly increased self-reported moderate physical activity from 97 minutes per week at enrollment to 325 minutes per week at 12 weeks, with no change observed among the controls. These data are consistent with previous findings from our team in a study of men with prostate cancer. In the Community of

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Wellness study, we observed a modest change in self-reported physical activity but only among men who reported <90 minutes per week of activity at baseline and in the group that received one coaching call with an exercise trainer [25]. It is possible that these previous studies reported greater changes in activity compared with this pilot study because they used self-report rather than objective measures. Nonetheless, some degree of coaching or more personalized contact may be needed to help people with cancer assess their current level of activity and identify what changes are needed to meet the physical activity guidelines and optimize their cancer outcomes.

Limitations

The baseline physical activity level measured at enrollment was high in both arms and particularly high in the intervention arm. This occurred despite the exclusion of prospective participants who self-reported ≥150 minutes per week of MVPA. However, physical activity measured using the accelerometers indicated that self-reported MVPA may have underestimated actual MVPA. It is also possible that participants engaged in higher than usual levels of activity when wearing the devices. Interestingly, when we analyzed moderate activity accumulated in bouts of ≥10 minutes, the participants' activity levels were similar to self-report. Although logistically difficult, future studies should consider using accelerometer data to determine eligibility or set a lower cutoff point for self-report to ensure they enroll an inactive study population who may most benefit from the intervention.

In addition, our sample included highly educated participants and low enrollment of Black or Latinx CRC survivors, which may limit the generalizability of our findings. Given the high CRC incidence and mortality among Black people and rising rates of young-onset CRC in some Latinx populations, research is critically needed in these patient groups [26,27]. Self-identified race or ethnicity was not assessed in our study until after participants provided consent. Although Hispanic or Latinx patients comprise 17% of patients with CRC at our institution, it is possible many may have been excluded owing to the requirement for English proficiency. We encourage future studies to support translations into multiple languages and to track the race or ethnicity of all screened participants to identify and address potential barriers to enrollment and ensure future studies enroll representative patient populations.

Finally, the COVID-19 pandemic began while the last 7 participants were active in the study. Several of our SMS text messages provided tips for participants to find social support and exercise with others, which were perceived by participants as irrelevant or incompatible with social distancing guidelines imposed during the pandemic. Out of these 7 participants, 6 (85%) participants had paired accelerometer data available. Out of these 6 participants, 5 (83%) decreased their time spent in bouts of moderate activity at 12 weeks compared with enrollment; 1 participant increased their time spent in moderate activity bouts. Although the numbers are small, it is possible that the pandemic led to a slightly greater decrease in planned moderate activity from enrollment to 12 weeks in our study, on average, than would have been observed in a study conducted before the COVID-19 pandemic. Decreases in physical activity, on average, have also been reported among noncancer study populations during the pandemic [28].

Future Work

Although the intervention was determined to be feasible and acceptable, there are aspects that could be improved in future studies. The main takeaway based on participant feedback was that the intervention, specifically the SMS text messages, needed to be more personalized. For example, several participants suggested that SMS text messages could be tailored to the data collected from the Fitbit. Future studies may be strengthened by having real-time access to activity data to determine appropriate messaging. Machine learning approaches, such as reinforcement learning, could also provide a platform to improve the tailoring of SMS text messages [29]. In addition, 4 participants in the intervention arm did not wear the Fitbit. Although the number is small, it is worth considering offering other mechanisms for self-monitoring physical activity, such as paper diaries, in future studies. Finally, studies are needed to determine the feasibility and acceptability of digital health physical activity interventions in individuals with lower levels of education, individuals with low English proficiency, and individuals who identify with minority racial or ethnic groups. Adaptation of digital health interventions and messaging into other languages and with attention to cultural contexts will be critical to improving access for a more diverse population.

Conclusions

Overall, this pilot study demonstrated that patients with CRC were interested in a remotely delivered, automated digital health physical activity intervention during chemotherapy. However, more tailored support is needed to further enhance participant satisfaction and possibly improve physical activity behavior.

Acknowledgments

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Conflicts of Interest

None declared.

Multimedia Appendix 1
Sample text messages from the Smart Pace II study.
[DOCX File , 26 KB - cancer_v8i1e31576_app1.docx]

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Multimedia Appendix 2
Participant feedback on the intervention components collected at 12 weeks using an open text write-in field in the participant feedback survey.
[DOCX File , 25 KB - cancer_v8i1e31576_app2.docx]

Multimedia Appendix 3
Mean physical activity, 6-minute walk test, body weight, and blood pressure at baseline and 12-week among participants in a 12-week pilot randomized controlled trial of a Fitbit Flex 2 and daily text messages.
[DOCX File , 27 KB - cancer_v8i1e31576_app3.docx]

Multimedia Appendix 4
CONSORT-eHEALTH checklist (V 1.6.1).
[PDF File (Adobe PDF File), 1154 KB - cancer_v8i1e31576_app4.pdf]

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Abbreviations

CHALLENGE: The Colon Health and Life-Long Exercise Change
CRC: colorectal cancer
FORCE: Focus on Reducing Dose-limiting Toxicities in Colon Cancer with Resistance Exercise
MVPA: moderate-to-vigorous physical activity
REDCap: Research Electronic Data Capture
UCSF: University of California, San Francisco

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Original Paper

Cruciferous Vegetable Intervention to Reduce the Risk of Cancer Recurrence in Non–Muscle-Invasive Bladder Cancer Survivors: Development Using a Systematic Process

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Abstract

Background: Bladder cancer is one of the top 10 most common cancers in the United States. Most bladder cancers (70%-80%) are diagnosed at early stages as non–muscle-invasive bladder cancer (NMIBC), which can be removed surgically. However, 50% to 80% of NMIBC cases recur within 5 years, and 15% to 30% progress with poor survival. Current treatments are limited and expensive. A wealth of preclinical and epidemiological evidence suggests that dietary isothiocyanates in cruciferous vegetables (Cruciferae) could be a novel, noninvasive, and cost-effective strategy to control NMIBC recurrence and progression.

Objective: The aim of this study is to develop a scalable dietary intervention that increases isothiocyanate exposure through Cruciferae intake in NMIBC survivors.

Methods: We worked with a community advisory board (N=8) to identify relevant factors, evidence-based behavior change techniques, and behavioral theory constructs used to increase Cruciferae intake in NMIBC survivors; use the PEN-3 Model focused on incorporating cultural factors salient to the group's shared experiences to review the intervention components (eg, the saliency of behavioral messages); administer the revised intervention to community partners for their feedback; and refine the intervention.

Results: We developed a multicomponent intervention for NMIBC survivors consisting of a magazine, tracking book, live telephone call script, and interactive voice messages. Entitled *POW-R Health: Power to Redefine Your Health*, the intervention incorporated findings from our adaptation process to ensure saliency to NMIBC survivors.

Conclusions: This is the first evidence-based, theoretically grounded dietary intervention developed to reduce bladder cancer recurrence in NMIBC survivors using a systematic process for community adaptation. This study provides a model for others who aim to develop behavioral, community-relevant interventions for cancer prevention and control with the overall goal of wide-scale implementation and dissemination.

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KEYWORDS

non–muscle invasive bladder cancer survivors; dietary intervention; cruciferous vegetable; cancer survivorship; cancer recurrence

Introduction

Background

Bladder cancer is the sixth most common cancer in the United States, contributing to >80,000 new cases and 17,000 deaths annually [1]. Bladder cancer mostly affects older adults, with an average age of diagnosis of 73 years. Men are 4 times more likely than women to be diagnosed with the disease, and the incidence rates in White men are double those of Black men [2]. Most bladder cancers (70%-80%) are diagnosed at early stages as non–muscle-invasive bladder cancer (NMIBC) [3]. After surgical removal, NMIBC frequently recurs (50%-80%), with some patients experiencing multiple recurrences at similar stages and others (15%-30%) progressing to muscle-invasive disease, which is associated with cystectomy and poor survival [3-5]. Novel, noninvasive, and cost-effective strategies to control NMIBC recurrence and progression are urgently needed.

Dietary isothiocyanates (ITCs) are phytochemicals primarily derived from cruciferous vegetables (Cruciferae, eg, kale, turnips, and broccoli) with multifaceted anticancer mechanisms [6,7]. ITCs are particularly promising against bladder cancer given that orally ingested ITCs are rapidly concentrated in urine and delivered to the bladder, maximizing direct exposure [8,9]. A compelling body of preclinical [8,10-12] and epidemiological evidence [13,14] has demonstrated the important role of dietary ITCs and ITC-rich Cruciferae in preventing bladder cancer recurrence and progression by inhibiting the growth of bladder cancer cells and preventing tumor progression. However, the consumption of the primary dietary source of ITCs—Cruciferae—is generally low among NMIBC survivors, with studies reporting 0.44-0.45 servings per day [13,15] and urinary ITC levels at approximately 4.4 µM, which is below the average of 10 µM of ITCs observed to inhibit ≥50% of bladder cancer cell growth in in vitro models [8,10].

There are currently no national guidelines regarding optimal Cruciferae intake to prevent bladder cancer recurrence in NMIBC. Urinary ITC levels are affected by the following: (1) ITC yield varies up to 300-fold from raw Cruciferae [16], (2) cooking reduces ITC yield [16], and (3) peak urinary concentration is achieved within 3 hours of dosing and >50% of the dose is excreted and accumulated in urine within 8 hours of dosing [11,17]. On the basis of these findings, we hypothesize that at least one serving (~1 cup raw or ½ cup cooked) of Cruciferae per day, with guidance on the choice of vegetables and cooking conditions, and consumption of Cruciferae at dinner time to minimize ITC excretion from frequent urination during the day will increase urinary ITC levels to the desired doses needed to exert anticancer activities in the bladder.

Objective

We describe a systematic process through which we developed an evidence-based Cruciferae intervention for NMIBC survivors with the goal of increasing Cruciferae intake in NMIBC survivors by at least one serving per day. The objectives of this study are to (1) describe how a well-known systematic process for evidence-based intervention adaptation can be used to develop a dietary intervention for cancer survivors and (2) detail an evidence-based, potentially scalable Cruciferae intervention that incorporates current preclinical and epidemiological data and NMIBC survivor perspectives. Our intention is that this study will facilitate future research that aims to improve NMIBC survivorship outcomes and inform the development process of future dietary interventions for cancer survivors.

Methods and Results

Overview

Consistent with the community-engaged approach of our study and other studies using a participatory process [18,19], we collaborated with an 8-member community advisory board to develop our intervention. We engaged our institution's networks to select community advisory board members that represented clinic staff, clinic providers, clinical research advocates, and NMIBC survivors, including an Asian male clinical urologist, a White female urology nurse, a Black cancer research advocate, 4 White male NMIBC survivors, and a White female NMIBC survivor. The constitution of the community advisory board represented both the local population and the diversity of patients with bladder cancer in terms of race and gender. Together, we went through a four-stage adaptation process [20] that consisted of (1) information gathering, (2) preliminary adaptation design, (3) preliminary adaptation tests, and (4) adaptation refinement (Multimedia Appendix 1). In stage 1, we collected data to inform intervention development. In stage 2, we decided on the skeleton of the intervention based on the data from stage 1. In stage 3, we developed a preliminary draft of the intervention. In stage 4, we refined the intervention. In this section, we report the methods and results of each of the 4 stages. The Institutional Review Board at Roswell Park approved all materials and methods.

Stage 1: Information Gathering

Stage 1 Methods

We collected data from the existing literature on dietary interventions for older adults, a discussion group with clinical staff, and in-depth interviews with NMIBC survivors.

Literature Review

The first author (KY) drew from the large body of literature on evidence-based behavioral fruit and vegetable interventions, focusing on interventions that significantly increased vegetable intake among participants whose demographic characteristics mirrored most NMIBC survivors (ie, aged ≥65 years and male) [21,22]. The search terms *dietary*, *diet*, *nutrition*, *vegetable*, *fruit*, *intervention*, and *review* were entered into PubMed to find review articles. Review articles were used to ascertain modalities (eg, telephone and in person) of intervention delivery that were associated with significant changes in dietary intake in older adults, and we chose the mode of intervention delivery based on this review. The first author then selected articles from the

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review papers that used our chosen intervention modality and caused significant changes in vegetable intake. From these studies, the first author, who is trained in qualitative methodology and behavioral interventions, identified the behavior change techniques incorporated into the interventions by using the behavior change technique taxonomy (version 1) [23], which identifies 93 distinct behavior change techniques. Behavioral theories used in these effective vegetable interventions were also examined to choose a theory to ground our intervention. After selecting the behavioral theory to ground the intervention, behavior change techniques that targeted the theory's constructs were included in the intervention's adaptation.

Discussion Group

After the literature review, we asked the clinical members of our community advisory board (ie, the urologist, nurse in urology clinic, and research advocate) to participate in a WebEx (Cisco Systems) discussion group to ascertain the factors necessary to maximize the saliency of an intervention for NMIBC survivors. Community members of the community advisory board (ie, NMIBC survivors) were not included in the discussion as we wanted to ascertain the shared experience of clinic members serving patients with NMIBC, which is a distinct experience from NMIBC survivors receiving treatment. During the discussion session, the facilitator (KY) asked discussion group members to draw from their experience working with NMIBC survivors to suggest topics to include in a Cruciferae intervention. The group was also asked what obstacles NMIBC survivors may have in eating more Cruciferae and about potential strategies to overcome any identified obstacles. The facilitator took notes during the discussion, summarized the main ideas from the discussion group, and recirculated the notes from the discussion to the discussion group members for verification.

In-depth Interviews

We then conducted in-depth interviews with NMIBC survivors. We used the PEN-3 Model [24,25] to develop the interview guide and analyze the data. The PEN-3 Model focuses on incorporating cultural factors salient to groups that share a similar collection of experiences [26,27]. Given that NMIBC survivors share a unique cancer treatment and control experience, we decided to use the PEN-3 Model to ensure that the intervention was salient to them (Multimedia Appendix 2) [24,25]. Analysis of the data consisted first of open coding followed by categorization of the codes based on PEN-3 dimensions.

We conducted in-depth interviews over the telephone with NMIBC survivor members of our community advisory board using a semistructured interview guide. The guide asked the participants to describe their experience with bladder cancer, their health goals and priorities, their knowledge of Cruciferae intake and cancer risk, barriers to and facilitators of higher Cruciferae intake, their food shopping habits, their preparation and planning of meals, and their social support for vegetable intake. All interviews were audio-recorded and transcribed.

A total of 2 research team members (KY and DE) began the PEN-3 process using an open coding technique in accordance with standard qualitative methodology [28,29]. Each research team member independently identified initial patterns, themes, and codes. For the *Cultural Identity* domain, the PEN dimensions of *Person*, *Extended Family*, and *Neighborhood* were considered in developing the codes. The 2 research team members then used the constant comparison approach to agree upon a list of final codes that emerged from the in-depth interview data. The finalized code list was then categorized by the dimensions of the *Relationships and Expectations* domain as *Perceptions*, *Enablers*, and *Nurturers*. We then used the *Cultural Empowerment* domain to determine whether the perceptions, enablers, and nurturers were *Positive*, *Existential*, or *Negative*. Any differences in categorization were resolved through discussion. The research team then used the analyzed data to develop intervention strategies.

Stage 1 Results

Literature Review

There were 6 review articles on dietary change interventions [30-35], of which 2 (33%) focused on dietary change in older adults [30,31]. The reviews reported that both face-to-face and telephone-based interventions had proven efficacy in changing dietary behavior in older adults [31,36]. Thus, to maximize our intervention's potential for scalability, we decided that our intervention would be telephone-based.

Of the 6 review articles, 4 (67%) used a telephone-based intervention and reported significant changes in vegetable intake in older adults (aged ≥60 years) [21,22,37,38]. The first author (KY) reviewed these 4 successful interventions and identified the application of the following evidence-based behavior change techniques [23]: problem solving or coping planning, social support (practical, general, and emotional), goal setting (outcome and behavior), prompts or cues, and feedback on behavior.

The 4 successful vegetable interventions reported the use of the transtheroretical model, health belief model, or social cognitive theory, or no theory at all [30,37]. There was no consensus regarding the intervention dosage or frequency of contact by intervention modality (eg, number of calls needed for dietary change).

Discussion Group

The discussion group included all clinical members of the community advisory board (3/8, 38%) and lasted approximately 60 minutes. Discussion group members reported that some of their patients with NMIBC called their bladder cancer a "fake cancer" because of their perception that it was a "just on the surface" cancer that had been successfully treated. These patients with NMIBC did not have a detailed understanding of how their cancer may progress and perceived the physician "scraping them" regularly as sufficient for long-term treatment. The NMIBC itself did not involve intensive treatment regimens such as chemotherapy or radiation therapy and, thus, was not seen as a dangerous disease. Consequently, discussion group members stated that the intervention needed to help the patient take NMIBC seriously without scaring them—that even though the risk of dying is low, the goal of the NMIBC survivor should be to prevent recurrence and save their bladder.

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Discussion group members also listed other potential obstacles to NMIBC survivors changing their dietary habits, including access to and cost of fresh Cruciferae, the intervention bringing back negative childhood memories of having to "eat their vegetables," and the physical discomfort (via gas) that could be caused by consuming Cruciferae. Despite the potential obstacles identified, discussion group participants stated that many of their patients asked them what they should eat and were eager to make dietary alterations to help fight the disease. They recommended ascertaining who did the grocery shopping for the NMIBC survivor as most survivors are older men who may not do their own food shopping.

In-depth Interviews

Project staff conducted 4 interviews with community advisory board members who were NMIBC survivors. Each interview lasted approximately 45 minutes. The NMIBC survivors included 3 non-Hispanic White men (3/4, 75%) and 1 non-Hispanic White woman (1/4, 25%). The mean age was 73 (SD 8.9) years, with 3 of them being married (3/4, 75%) and 1 retired (1/4, 25%). Of the 4 participants, 2 (50%) reported completion of high school as their highest level of education, whereas 1 (25%) reported at least some college education, and 1 (25%) reported postgraduate education. All participants (4/4, 100%) had health insurance, with 3 reporting Medicare (3/4, 75%) and 1 reporting Medicaid (1/4, 25%). Multimedia Appendix 3 presents the results of the PEN-3 analysis according to the *Relationships and Expectations* domain of perceptions, enablers, and nurturers. Some of the codes are listed more than once if they covered >1 PEN-3 dimension. Within each domain, data were then categorized by the *Cultural Empowerment* domain dimensions of positive, existential, and negative.

Perceptions

Positive

NMIBC survivors' positive perceptions included knowledge regarding the benefits of healthy eating, particularly vegetable intake. Some participants spoke about the daily routine of having no choice but to eat what was on their plate—including fresh vegetables—in the "previous generation" when they were children. NMIBC survivors believed that fresh vegetables were best and that information from physicians and others in authority had more validity.

Existential

Many NMIBC survivors described their initial diagnosis of bladder cancer as a shock. Some described no symptoms before diagnosis and described the treatment as minimal. Many participants also said they were unaware of the evidence linking Cruciferae with decreased bladder cancer recurrence.

Negative

Negative perceptions included seeing bladder cancer as a "lesser" or "good" cancer and the sufficiency of maintenance medical visits to prevent bladder cancer recurrence. Many NMIBC survivors said they felt well and wanted to maintain a positive attitude; consequently, they were unaware of the high risk of recurrence of bladder cancer. Regarding the consumption of Cruciferae, participants reported the flavor, texture, appearance, and effort required for preparation as barriers. Overcoming habits of poorer eating choices was also a negative perception.

Enablers

Positive

All the NMIBC survivors emphasized the wife's strong role in promoting healthy dietary behaviors in survivors. Other positive enablers included perceived easy access to a variety of fresh vegetables and vegetable recipes. Many survivors reported that their bladder cancer diagnosis was a catalyst for eating healthier.

Existential

Some NMIBC survivors described difficulty in acknowledging that they were cancer survivors as their cancer treatment resulted in minimal side effects.

Negative

Negative enablers included feelings of guilt for being considered a cancer survivor because of the less intensive treatments they underwent compared with other cancer survivors. Consequently, some did not consider bladder cancer a "real" cancer for them to be vigilant in preventing. The time and effort required to prepare Cruciferae, friends and fellow survivors' poor dietary habits, and television advertisements for less healthy food choices were other identified negative enablers. The medical system's provision of ongoing monitoring after treatment also served as a negative enabler for dietary change as medical management alone was perceived as enough to prevent recurrence.

Nurturers

Positive

NMIBC survivors overwhelmingly discussed the survivor's wife as a positive nurturer who set nutritional priorities, prepared meals, and went grocery shopping. Physicians were also identified as positive nurturers to convince survivors of the importance of Cruciferae.

Existential

There were no existential nurturers identified.

Negative

Negative nurturers included having family members with poor health who required care and took away from the survivors' ability to care for themselves and the lack of a wife, which would make healthy food preparation tasks insurmountable to the survivor.

Stage 2: Preliminary Adaptation Design

Stage 2 Methods

We performed an iterative process to incorporate the results from stage 1 to develop preliminary drafts of our Cruciferae intervention for NMIBC survivors. The results from stage 1 were discussed as a research team and with our community advisory board. After we came to a consensus concerning how to include the results from stage 1, a subgroup within our team developed an initial draft of the intervention, which was then recirculated back to the larger group for feedback and subsequent refinement.

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Stage 2 Results

Literature Review

Data from stage 1 were used to develop the basic framework for our intervention. We decided to incorporate all the evidence-based behavior change techniques [23] used by the 4 papers [21,22,37,38] that described phone-based interventions significantly increasing vegetable intake in older adults: problem solving or coping planning, social support (practical, general, and emotional), goal setting (outcome and behavior), prompts or cues, and feedback on behavior. We incorporated these behavior change techniques throughout the intervention, for example, we included an action plan for each participant to complete to set short- and long-term Cruciferae intake goals (goal setting) and a process for the participants to receive ongoing feedback regarding their Cruciferae intake through interactive voice response (IVR), which is described below (feedback on behavior). Given that dietary interventions grounded in theory may be more effective than non–theoretically-based interventions [30], we decided to base our intervention on theory. We chose social cognitive theory as the evidence-based behavior change techniques identified in our review were linked to 8 of the 11 major social cognitive theory constructs (self-efficacy; outcome expectations; knowledge; social support; barriers and opportunities; behavioral skills; intentions; and reinforcement and punishments). Thus, these constructs were interwoven throughout our intervention (eg, intervention components were designed to boost self-efficacy, knowledge, and specific behavioral skills by incorporating information about Cruciferae and strategies to increase intake). The 3 major constructs of social cognitive theory that were not used in the 4 studies reviewed (collective efficacy, observational learning, and normative beliefs) were included by adding behavior change techniques (restructuring the social environment, credible source, and social comparison) that reflected these constructs.

We chose a telephone-based modality for intervention delivery as this method has proven efficacy in changing dietary behavior in older adults [31] and would arguably be more feasible to implement on a wider scale compared with face-to-face interventions [36]. We also decided to deliver the bulk of our telephone calls through automated calls or IVR telephone messages to facilitate future scale-up. Given the lack of consensus regarding intervention dosage or frequency of contact by intervention modality, we decided to include 1 mailing, 1 live telephone call, and 11 IVR calls over a 6-month period. We adapted an IVR template with proven success in a previous dietary change intervention [39] to develop our IVR calls.

Thus, the structure of our intervention consisted of (1) an initial mailing of an informational magazine and booklets to track Cruciferae intake; (2) a follow-up telephone call with research staff to verify understanding of the educational information, provide instructions on how to use the track books, and help participants complete a personalized action plan whereby participants identified barriers and facilitators to meet identified goals; and (3) 11 IVR telephone calls whereby participants entered the amount of Cruciferae consumed and received tailored feedback based on their reported consumption.

Specific content within our intervention was informed through the results of the discussion group and in-depth interviews conducted in stage 1.

Discussion Group

We specifically addressed the idea that NMIBC is perceived as a fake cancer by emphasizing the importance of taking the cancer seriously. Potential obstacles to Cruciferae intake identified by the discussion group were also included in the IVR specifications. Identifying who in the household did the grocery shopping and ensuring that they were engaged in the participant's action plan was also included.

In-depth Interviews

We developed intervention strategies based on the categories of the PEN-3 framework according to which the qualitative data were sorted. Specifically, the intervention strategies were based on the perception, enabling, and nurturing dimension within the *Relationships and Expectations* domain. Intervention strategies either reinforced factors that supported increased Cruciferae intake or revised factors that discouraged dietary change to facilitators of dietary change. The specific strategies, sorted by reinforcing or revised strategies, are presented in Multimedia Appendix 4. Overall, the strategies emphasized the high recurrence rate among NMIBC survivors and the fact that medical monitoring alone will not prevent recurrence. Identified strategies included presenting information about significant relationships between Cruciferae intake and bladder cancer etiology, the role of Cruciferae in staying strong and living long, easy ways to prepare Cruciferae, and an emphasis on fresh vegetable consumption. Engaging spouses or partners, family, and friends to change their dietary behavior was also promoted. Given the survivors' lack of saliency with the word *cruciferous*, the intervention was branded as the *Power to Redefine Your Health* (POW-R Health) Program, wherein Cruciferae are referred to as *power* vegetables. The POW-R Health Program is promoted as an intervention developed and endorsed by medical professionals.

Stage 3: Preliminary Adaptation Tests

Stage 3 Methods

On the basis of the data collected in stages 1 and 2, we developed draft educational materials, a script for the live call, and the IVR specifications. Our team then engaged in an iterative process to ensure that the intervention was salient to NMIBC survivors. The initial drafts of the magazine, track books, and IVR specifications were reviewed by the team. The team interventionist pilot-tested the live call script with 3 NMIBC survivors in the community advisory board.

Stage 3 Results

Team members suggested that a project logo be developed to promote continuity between the various pieces of the intervention. Thus, we worked with the NMIBC survivors in the community advisory board to develop a project logo. NMIBC survivors reviewed various logos and chose a circular one with pictures of Cruciferae. The logo was included in the educational materials (magazine and track book).

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Some team members reported that directly stating in the intervention materials that 50% to 80% of NMIBC recurs within 5 years may scare survivors and recommended removing the statistic. Thus, we developed different versions to convey the high risk of bladder cancer recurrence and presented these versions to the NMIBC survivors in the community advisory board. The NMIBC survivors agreed that the high risk of recurrence needed to be directly conveyed by reporting the statistic. Several NMIBC survivors stated that "telling the truth as it is" would not unnecessarily scare survivors but rather emphasize the urgency to care for one's health.

A larger font size (14 points) was recommended to make the magazine easier to read in addition to pictures showing diverse older adults. The team decided to add to the track book cover the total cups of Cruciferae consumed throughout the week to facilitate participant entry of that information for the IVR calls. The community advisory board thought that the track book was easy to understand and use.

All recommended that the initial 60-minute telephone call be shortened; thus, the initial portion of the call that included an in-depth overview of the magazine was changed from being delivered to every participant to being delivered only to those participants who did not review the magazine before the telephone call. The NMIBC survivors emphasized the importance of delivering the live call with energy and in a conversational manner.

Finally, the team recommended the creation of a magnet to remind the participants of the project IVR telephone number. In addition, the IVR number was highlighted at the end of the magazine, on the back page of the track book, and at the end of the live call to facilitate IVR use.

Stage 4: Adaptation Refinement

Stage 4 Methods

Data from stages 1-3 were used to develop the near-final versions of the POW-R Health intervention, with these versions reviewed by the entire community advisory board and investigative team before finalization.

Stage 4 Results

Minor editorial adjustments were made to yield the final product—a Cruciferae intervention for NMIBC survivors designed to increase Cruciferae intake by at least one serving a day to reduce the risk of bladder cancer recurrence. The intervention consists of (1) an 11-page, 21.6 cm by 27.9 cm color magazine that includes information about the high risk of bladder cancer recurrence in NMIBC survivors, what Cruciferae are, and strategies to maximize ITC yield from Cruciferae consumption (≥1 cup a day at or after dinner, eaten raw or lightly cooked, of select high– to medium–ITC-yield Cruciferae) as well as an action plan with short- and long-term goal setting, identified obstacles, and strategies to meet set goals; (2) weekly track books to monitor Cruciferae intake throughout the duration of the 6-month intervention to facilitate self-monitoring; (3) a 45-minute live telephone call following receipt of the magazine and track books with an interventionist who ensures that the participants understand the materials sent, completes the action plan with the participants, and practices how to fill out the track books; and (4) 11 IVR calls after the live call spread across the remainder of the 6-month intervention whereby the participants enter the amount of Cruciferae consumed in cups and the IVR provides feedback based on the participants' previously stated goals (eg, *Mark, I am really pleased to see that you accomplished your goal for the week! Congratulations on your success! The changes you are making are really going to help with your health in the long run*...or *Frank, based on what you ate in total last week, it looks like you didn't quite hit your goal, but you did make some progress so congratulations on your success! Lots of people struggle with trying to eat more power vegetables*...). IVR calls are made automatically to the participants, and the participants can call into the IVR system if they miss an IVR call. A magnet with the project logo and IVR telephone number is given to the participants to maximize their IVR use.

Discussion

Principal Findings

We used a systematic approach to integrate the most up-to-date scientific knowledge of bladder cancer etiology, evidence-based behavior change techniques in dietary interventions, behavioral theory, and community expertise to develop POW-R Health. We believe that this systematic approach will maximize the intervention's potential to produce clinically meaningful change [40]. We sought to produce an intervention salient to NMIBC survivors by making NMIBC survivors' experiences, perceptions, behaviors, and knowledge the focal point of our process. Our choice of using telephone-based modalities and minimal staff contact also arguably increases the intervention's scalability potential.

Compared with the development process of other dietary interventions for cancer survivors, our systematic approach for developing POW-R Health involves greater community engagement (eg, via the community advisory board) and the inclusion of more informational sources to inform intervention development. Most dietary interventions for cancer survivors have been developed without community input and created solely by the academic or clinical team [41,42]. Among the few that engaged the community, focus groups and surveys were used to garner feedback [43-46]. A total of 3 interventions used components from previous evidence-based interventions to develop their intervention [47-49]. Similar to our process, most published dietary interventions for cancer survivors were grounded in behavioral theory [41,42]. One study mirrored our process of including a literature review and choosing behavior change techniques to develop their intervention [43].

This study is not without limitations. First, members of the community advisory board did not include racially and ethnically diverse NMIBC survivors, although we attempted to mitigate this to some degree by including a diverse clinical team, among them a Black community cancer research advocate and cancer survivor. The resulting intervention model is expected to match well with the demographics of most NMIBC survivors and can provide a basis for further demographic tailoring in additional versions. Second, our literature review

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did not include multiple reviewers, which would have increased the rigor of the methods. Third, although many recurring themes emerged from the in-depth interviews with NMIBC survivors, we acknowledge that complete data saturation may not have been achieved. Acknowledging these limitations, we plan to conduct in-depth exit interviews at the end of the intervention study with all the participants to ascertain relevant aspects from more NMIBC survivors. These limitations should be considered within the strengths of this study, including collaboration with a community advisory board to guide all stages of this systematic intervention adaptation process and the first known evidence-based dietary intervention developed specifically for NMIBC survivors to reduce bladder cancer recurrence.

Currently, POW-R Health is being compared with an alternative treatment control in a pilot randomized controlled trial with NMIBC survivors. We hypothesize that, compared to the control, POW-R Health will significantly increase Cruciferae intake and urinary ITC levels and alter gene expression associated with bladder cancer recurrence. For cancer researchers who aim to develop behavioral interventions for cancer survivors, we encourage the use of similar systematic evidence-based frameworks that meaningfully engage communities in addition to evidence-based behavior change techniques and behavioral theory.

Conclusions

Bladder cancer is a serious disease in which up to 80% of NMIBC survivors experience recurrence. Most patients with NMIBC have no treatment offered to prevent recurrence, instead relying on costly and frequent surveillance to monitor disease status with the goal of capturing recurrence or progression at early stages of treatment. Our intervention offers the potential for a promising, feasible, and accessible way for all NMIBC survivors to reduce bladder cancer recurrence. Our hope is that, if POW-R Health is proven to be efficacious, Cruciferae intake recommendations will become a part of standard clinical practice and meaningfully reduce negative outcomes associated with NMIBC.

Acknowledgments

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Conflicts of Interest

None declared.

Multimedia Appendix 1
The 4-stage adaptation process for intervention development.
[PPTX File , 52 KB - cancer_v8i1e32291_app1.pptx]

Multimedia Appendix 2
PEN-3 Model.
[DOCX File , 12 KB - cancer_v8i1e32291_app2.docx]

Multimedia Appendix 3
Summary of qualitative results according to the PEN-3 framework.
[DOCX File , 18 KB - cancer_v8i1e32291_app3.docx]

Multimedia Appendix 4
Intervention strategies categorized by PEN-3 dimensions.
[DOCX File , 17 KB - cancer_v8i1e32291_app4.docx]

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Abbreviations

ITC: isothiocyanate
IVR: interactive voice response
NMIBC: non–muscle-invasive bladder cancer
POW-R Health: *Power to Redefine Your Health*

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Original Paper

A Web- and Mobile-Based Intervention for Women Treated for Breast Cancer to Manage Chronic Pain and Symptoms Related to Lymphedema: Results of a Randomized Clinical Trial

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Abstract

Background: The-Optimal-Lymph-Flow (TOLF) is a patient-centered, web- and mobile-based mHealth system that delivers safe, easy, and feasible digital therapy of lymphatic exercises and limb mobility exercises.

Objective: The purpose of this randomized clinical trial (RCT) was to evaluate the effectiveness of the web- and mobile-based TOLF system for managing chronic pain and symptoms related to lymphedema. The primary outcome includes pain reduction, and the secondary outcomes focus on symptom relief, limb volume difference measured by infrared perometer, BMI, and quality of life (QOL) related to pain. We hypothesized that participants in the intervention group would have improved pain and symptom experiences, limb volume difference, BMI, and QOL.

Methods: A parallel RCT with a control–experimental, pre- and posttest, and repeated-measures design were used. A total of 120 patients were recruited face-to-face at the point of care during clinical visits. Patients were randomized according to pain in a 1:1 ratio into either the arm precaution (AP) control group to improve limb mobility and arm protection or The-Optimal-Lymph flow (TOLF) intervention group to promote lymph flow and limb mobility. Trial outcomes were evaluated at baseline and at week 12 after the intervention. Descriptive statistics, Fisher exact tests, Wilcoxon rank-sum tests, t test, and generalized linear mixed effects models were performed for data analysis.

Results: At the study endpoint of 12 weeks, significantly fewer patients in the TOLF intervention group compared with the AP control group reported chronic pain (45% [27/60] vs 70% [42/60]; odds ratio [OR] 0.39, 95% CI 0.17-0.90; *P*=.02). Patients who received the TOLF intervention were significantly more likely to achieve a complete reduction in pain (50% [23/46] vs 22% [11/51]; OR 3.56, 95% CI 1.39-9.76; *P*=.005) and soreness (43% [21/49] vs 22% [11/51]; OR 2.60, 95% CI 1.03-6.81; *P*=.03). Significantly lower median severity scores were found in the TOLF group for chronic pain (MedTOLF=0, IQR 0-1 vs MedAP=1,

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IQR 0-2; *P*=.02) and general bodily pain (MedTOLF=1, IQR=0-1.5 vs MedAP=1, IQR 1-3; *P*=.04). Compared with the AP control group, significantly fewer patients in the TOLF group reported arm/hand swelling (*P*=.04), heaviness (*P*=.03), redness (*P*=.03), and limited movement in shoulder (*P*=.02) and arm (*P*=.03). No significant differences between the TOLF and AP groups were found in complete reduction of aching (*P*=.12) and tenderness (*P*=.65), mean numbers of lymphedema symptom reported (*P*=.11), ≥5% limb volume differences (*P*=.48), and BMI (*P*=.12).

Conclusions: The TOLF intervention had significant benefits for breast cancer survivors to manage chronic pain, soreness, general bodily pain, arm/hand swelling, heaviness, and impaired limb mobility. The intervention resulted in a 13% reduction (from 40% [24/60] to 27% [16/60]) in proportions of patients who took pain medications compared with the AP control group, which had a 5% increase (from 40% [24/60] to 45% [27/60]). A 12% reduction (from 27% [16/60] to 15% [9/60]) in proportions of patients with ≥5% limb volume differences was found in the TOLF intervention, while a 5% increase in the AP control group (from 40% [24/60] to 45% [27/60]) was found. In conclusion, the TOLF intervention can be a better choice for breast cancer survivors to reduce chronic pain and limb volume.

Trial Registration: Clinicaltrials.gov NCT02462226; https://clinicaltrials.gov/ct2/show/NCT02462226

International Registered Report Identifier (IRRID): RR2-10.2196/resprot.5104

(JMIR Cancer 2022;8(1):e29485) doi:10.2196/29485

KEYWORDS

pain; lymphatic exercises; symptoms; lymphedema; breast cancer; health behavior; mHealth

Introduction

Background

Annually, more than 260,000 women are diagnosed with breast cancer, and currently there are more than 3.8 million breast cancer survivors in the United States [1]. Even years after cancer treatment, about 40% of women treated for breast cancer suffer daily from chronic pain and more than 50% of women report multiple distressing symptoms related to lymph fluid accumulation [2-5]. The abnormal accumulation of lymph fluid after breast cancer treatment is a result of obstruction or disruption of the lymphatic system associated with cancer treatment (eg, removal of lymph nodes or radiotherapy), influenced by patient personal factors (eg, obesity or higher BMI), and triggered by factors such as infections or trauma [6-8]. The accumulation of lymph fluid leads to chronic and various pain sensations (ie, pain/aching/soreness/tenderness) in the ipsilateral upper limb or body and other symptoms related to fluid accumulation defined as lymphedema symptoms [3,9].

While significantly more breast cancer survivors with a diagnosis of lymphedema experience pain (45.2%), tenderness (52.4%), aching (61.9%), or soreness (31%), a substantial amount of breast cancer survivors without a diagnosis of lymphedema also experience pain (40%), tenderness (47.3%), aching (30%), or soreness (32.7%) [9]. On average, breast cancer survivors without lymphedema report about 5 lymphedema symptoms while breast cancer survivors with lymphedema report 10 symptoms [9,10]. Despite current advances in cancer treatment, it is clear that many breast cancer survivors still face long-term postoperative challenges as a result of experiencing daily pain and lymphedema symptoms.

Pain and lymphedema symptoms are debilitating late complications that impact the breast cancer survivors' quality of life (QOL) [2,3,5,11]. Persistent pain related to cancer treatment is considered a stressful complication because it is perceived as a constant reminder of cancer [2,12] and exerts tremendous limitations on breast cancer survivors' daily living [2,5]. Pain and lymphedema symptoms can instigate fears and induce feelings of loss of control [2,3,5]. Specifically, the experience of pain, including tenderness, aching, or soreness, causes significant and unrelenting distress among breast cancer survivors [3]. Such distress is usually heightened when breast cancer survivors expect pain and symptoms related to lymphedema to disappear but instead stay as a "perpetual discomfort" [3] (p853). The negative impact of pain and lymphedema symptoms can be a source of considerable disability and psychological distress that negatively influences the patient's daily living [2,3,11,12] and creates a tremendous burden on the health care system [13]. Nonetheless, in clinical practice pain and symptoms related to lymphedema are still underrecognized and undertreated.

While more research is needed to explore the exact etiology of persistent pain and lymphedema symptoms (eg, arm swelling, breast swelling, chest wall swelling, heaviness, firmness, tightness, stiffness, numbness, burning, stabbing, tingling, and limited limb movement), physiologically, the accumulation of lymph fluid in the affected area or limb may create undue pressure on nerves, producing feelings of pain, aching, tenderness, soreness, burning, tingling, stabbing, and numbness as well as inducing sensations of swelling, heaviness, tightness, and firmness [14,15]. Accumulated lymph fluid in the affected area or limb also leads to stiffness and limited limb movement of the arm, shoulder, fingers, and elbow [10,15]. Significant associations are found between pain (including aching and tenderness) and accumulation of lymph fluid in the ipsilateral upper limb [10,15]. Research has also shown that with the increased number of symptoms reported, breast cancer survivors' limb volume increased [10,15]. Limb volume as detected by the infrared perometer has significantly elevated as breast cancer survivors' reports of pain, tenderness, aching, swelling, heaviness, firmness, and tightness have increased [10]. For breast cancer survivors without a diagnosis of lymphedema, persistent pain and lymphedema symptoms are cardinal symptoms of early stage lymphedema because such symptoms often precede changes in limb size or girth or a lymphedema

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diagnosis [9]. Without a timely intervention, this early disease stage can progress into lymphedema that no surgical or medical interventions can cure [7,15].

Breast cancer survivors are known to have a compromised lymphatic system due to breast surgery, dissection of lymph nodes and vessels, and radiation, which leads to ineffective lymphatic drainage, thus accumulation of lymph fluid in the affected area or limb [10,15,16]. In addition to the risk factor of compromised lymphatic drainage from cancer treatment, higher BMI is also an established risk factor for the accumulation of lymph fluid [6-10]. Physiologically, a larger body mass creates a disproportion in lymph transport and capacity, resulting in excess extracellular fluid [6,17]. Women are 1.11 times more at risk for developing lymphedema with every 1 kg/m^2 increase in their BMI [6-8,16]. Although the known risk factors for symptoms related to accumulation of lymph fluid directly from cancer treatment cannot be avoided (such as removal of lymph nodes, surgery, radiation, chemotherapy, and hormonal therapy), some risk factors (such as compromised lymphatic drainage and higher BMI) can be modified through education and self-care strategies [14,18,19].

Patient education focusing on self-care strategies holds great promise for reducing the risk of lymph fluid accumulation [14,18,19]. Research evidence demonstrates that even after controlling for confounding cancer treatment–related risk factors, patient education on self-care strategies remains an important predictor for patient-centered outcomes, including symptom experience and self-care behaviors [14,18,19]. Current patient education emphasizes precautionary lifestyle behaviors, such as avoidance of repetitive limb movement, lifting weighted objects, needle punctures, blood draw, and the use of compression garments for air travel in the affected limb [20,21]. To date, there is a paucity of high-quality evidence to support these precautionary practices that reduce the risk of lymphedema and relieve pain or symptoms related to lymph fluid accumulation [20,21]. Research is lacking to provide evidence to reduce pain and symptoms related to lymph fluid accumulation through self-care strategies targeting compromised lymphatic drainage and higher BMI.

Grounded in research-driven self-care behavioral strategies [14,19], The-Optimal-Lymph-Flow (TOLF) [22], a unique patient-centered web- and mobile-based educational and behavioral program, focuses on self-care strategies targeting compromised lymphatic system to promote lymph flow, limb mobility, and maintaining optimal BMI, that is, risk factors for pain and lymphedema symptoms. Patients learn self-care strategies through the web- and mobile-based program that can be downloaded on a computer, laptop, and any mobile phones and tablets. Its underlying premise is to empower, rather than inhibit, how breast cancer survivors live their lives by emphasizing “what to do,” rather than “what to avoid.” It features a safe, feasible, and easily integrated-into-daily-routine self-care strategies that include therapeutic lymphatic exercises (ie, muscle tightening–breathing, muscle tightening–pumping exercises, and large muscle exercises) to promote lymph flow and drainage, limb mobility exercises to promote shoulder and arm function, and general instructions to encourage nutrition-balanced (more vegetables and fruits), portion-appropriate diet (feeling 75% full for each meal), adequate hydration, and sleep to strive for maintaining optimal BMI. Patients can learn and follow all the exercises through avatar video simulations [14,19]. The efficacy of The Optimal Lymph-Flow has been demonstrated in our recently published study of 140 patients who received the face-to-face nurse-delivered program [19]. Findings of the study demonstrated that over 90% of patients improved their limb volume at the 12-month follow-up. This system has been used successfully for its usability testing. The preliminary usability and feasibility testing were completed with 30 breast cancer survivors who evaluated the easiness, difficulties, and feasibility of using the system on computer, iPhone, iPad, or other smartphones or tablets [23]. Findings of the usability and feasibility testing have demonstrated that patients love the web-based program, especially the videos using the avatar technology to demonstrate the complicated lymphatic system, and illustrate the physiological functions of each exercise and detailed step-by-step instructions for each exercise.

Objectives and Hypotheses

The purpose of this randomized clinical trial (RCT) was to evaluate the efficacy of the web- and mobile-based TOLF system, a patient-centered educational and behavioral symptom management program focusing on promoting lymph flow, improving limb mobility, and optimizing BMI, for managing chronic pain and lymphedema symptoms.

The primary objective of this study was to determine the effectiveness of the web- and mobile-based TOLF system for managing chronic pain, aching, soreness, tenderness, and general bodily pain among breast cancer survivors. We hypothesized that more patients who received the TOLF intervention would report a complete reduction and reduced severity of pain, aching, soreness, tenderness, and general bodily pain compared with patients who received the arm precaution (AP) control at week 12 after the intervention.

The secondary objective of the study was to evaluate the effectiveness of the web- and mobile-based TOLF system for managing lymphedema symptoms, limb volume differences, BMI, and QOL related to pain. We hypothesized that patients who received the TOLF intervention would report fewer lymphedema symptoms, minimal limb volume differences, and better BMI and QOL compared with patients who received the AP control.

Methods

Ethical Approval

This study (IRB# i15-00221) was approved by the Institutional Review Board of New York University Langone Medical Center on June 8, 2015.

Design

Chronic pain, including aching, soreness, and tenderness, is defined as persistent or intermittent pain in the ipsilateral upper limb or body at least 3 months after surgical treatment for breast cancer, that is, beyond the expected period of healing [24,25].

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A 12-week, 2-arm, parallel RCT (ClinicalTrials.gov Identifier: NCT02462226) was designed to evaluate the effectiveness of the web- and mobile-based TOLF self-care strategies to promote lymph flow versus control AP for managing chronic pain and lymphedema symptoms. The data collectors were blinded to the group assignments. The protocol was in accordance with the CONSORT-EHEALTH checklist (Multimedia Appendix 1) [26].

Setting

The study was conducted in a nursing research laboratory located in the breast cancer clinic of New York University Laura and Isaac Perlmutter Cancer Center, a National Cancer Institute–designated cancer center in New York City.

Study Participants

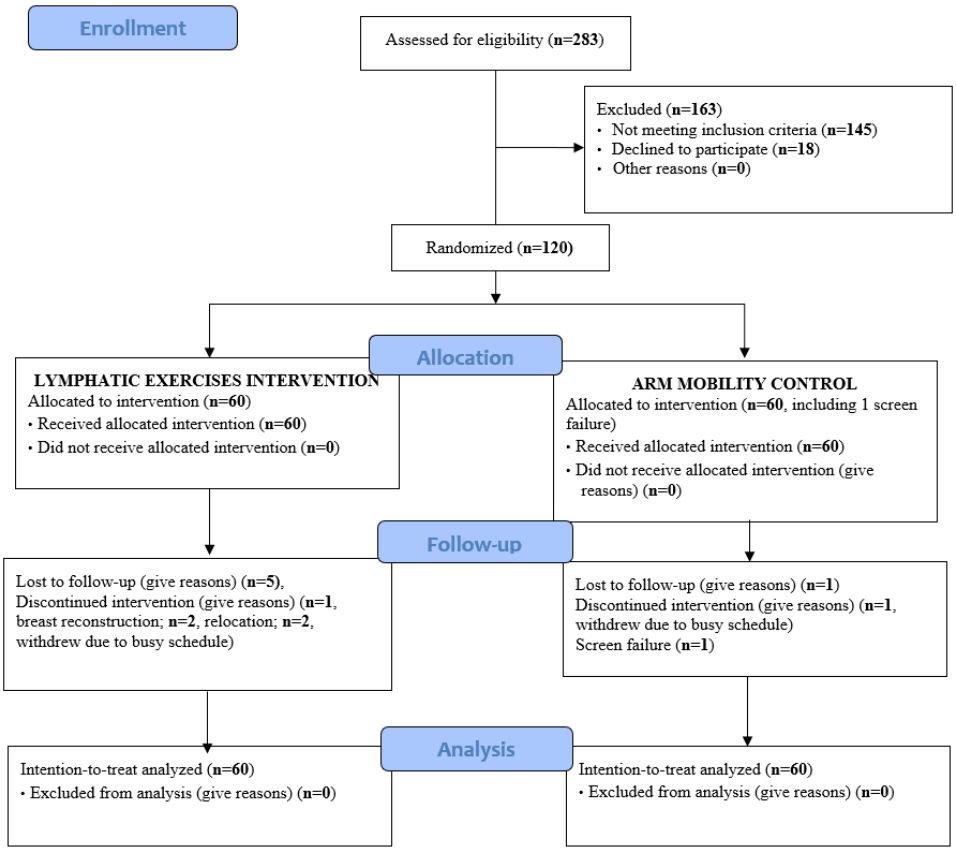
Study participants included (1) patients who received surgical treatment for cancer at least 3 months prior to the study enrollment, because healing usually occurs within 3 months of surgical treatment for cancer [24,25]; (2) patients who reported *persistent or intermittent* pain (including aching, soreness, or tenderness); (3) patients who may or may not report any of lymphedema symptoms (ie, swelling, heaviness, tightness, firmness, numbness, tingling, stiffness, limb fatigue, limb weakness, and impaired limb mobility of shoulder, arm, elbow, wrist, and fingers); (4) patients who may or may not have a history of lymphedema or have been treated for lymphedema; (5) patients who had internet access to the web- and mobile-based program at home or were willing to access the program using the laptop provided by the researchers at the cancer center; (6) patients who had the ability to understand and the willingness to sign a written informed consent document.

Exclusion criteria were (1) patients who did not report any pain, including aching, soreness, or tenderness; (2) patients who had a known metastatic disease or other bulk disease in the thoracic or cervical regions; (3) patients who had lymphedema due to cancer recurrence; and (4) patients who had documented advanced cardiac or renal disease.

Recruitment

From June 17, 2015, to December 1, 2016, we screened 283 patients for eligibility and enrolled and randomized 120 patients and followed the participants for 12 weeks after the intervention. Among the 283 patients screened, 163 were excluded for the following reasons: (1) not meeting inclusion criteria (n=145) and (2) declined to participate (n=18). Participants were recruited face to face at the point of care during clinical visits from the New York University Perlmutter Cancer Center. Figure 1 presents the CONSORT-EHEALTH diagram for recruitment and randomization.

Figure 1. CONSORT-EHEALTH flowchart for recruitment.



To accomplish the recruitment of 120 participants, we used the successful procedures of recruiting and consenting participants used by the principal investigator and the team in the preliminary studies [3,9,14,17,19]. Successful strategies included the use of *invitation flyers* that described the study. This *invitation flyer* was posted on the bulletin boards or breast cancer support website at the cancer center, and was also available in the reception areas of the cancer center, examination rooms, and rooms holding support group meetings.

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Consent Process

After reading the *invitation flyer*, women who were interested in participating in the study called and scheduled a meeting with the research coordinator. During the meeting, the research coordinator confirmed the woman's interest, determined if the woman was eligible for the study, and the research coordinator again explained the study in detail and provided enough time for the woman to ask questions. Protection of human participants was ensured by following the guidelines set forth by the Institutional Review Board. Each participant signed the written study consent.

Randomization and Blinding

The randomization assignment was generated by our senior statistician (GY) using a computer-generated randomization procedure. Participants were randomized based on their report of pain/aching/soreness or tenderness to be allocated with a 1:1 ratio to either the TOLF intervention or the AP control group. The researchers who performed pre- and postintervention measurements were blinded throughout the study to the participants' assigned arm. Participants did not know which intervention was the intervention of interest and which one was the comparator. Of the 120 patients enrolled, 60 were assigned to the TOLF intervention group and 60 to the AP control group (Figure 1).

Study Intervention

Overview

The web- and mobile-based TOLF system [19,22,23] included information about lymphedema, diagnosis and measurement of lymphedema, lymphatic system, risk of lymphedema, self-care, daily therapeutic exercises, APs, and Ask Experts. Participants in the TOLF intervention group had access to the 8 avatar videos that provided step-by-step instructions for TOLF lymphatic exercises to promote lymph flow and optimize shoulder and limb mobility. The platform also has a section entitled *Arm Precautions*, representing current patient education that emphasizes precautionary lifestyle behaviors, such as avoidance of repetitive limb movement, lifting weighted objects, needle punctures, blood draw, and the use of compression garments for air travel in the affected limb [20,21]. Table 1 presents the strategies, rationales, and actions for the TOLF intervention.

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Table 1. The-Optimal-Lymph-Flow Program: self-care strategies, rationales, and actions.

Strategies and exercises	Rationales	Actions
Promoting lymph flow		
• Muscle tightening deep breathing	• The whole-body lymph fluid has to be drained through the lymphatic ducts above the heart. Muscle tightening–deep breathing stimulates lymphatic ducts and helps lymph fluid drain. • Lymph fluid drains when muscles move. Muscle tightening–deep breathing creates the whole-body muscle movements that create muscle milking and pumping action and help to drain lymph fluid.	• At least twice a day in the morning and at night before brushing teeth or as much as the patient wants throughout the day. • Air travel: before take-off and after landing. • Sedentary lifestyle: At least every 4 hours.
• Muscle tightening–pumping	• Muscle tightening–pumping exercises create arm muscle pumping. This helps lymph fluid flow and decreases the fluid build-up in the arms. • Muscle tightening–pumping exercises build the arm muscle that helps lymph fluid flow and drain.	• At least twice a day in the morning and at night before brushing teeth or as much as the patient wants throughout the day. • Air travel: before take-off and after landing. • Sedentary lifestyle: At least every 4 hours.
Improving limb mobility		
• TOLF[a] limb mobility exercises: shoulder rolls, clasp and spread, and reach to the sky. • Arm precaution limb mobility exercises: shoulder rolls, clasp and spread, reach to the sky, wall climb, and sideway wall stretches.	• Improved limb mobility after surgery facilitates local muscle movements that create muscle milking and pumping to promote local limb lymph fluid flow and drain. • Shoulder exercises create arm muscle milking and pumping by moving the main anterior upper arm muscles (biceps brachii, brachialis, coracobrachialis), the posterior muscle of triceps brachii, and deltoid muscle (ie, the anterior deltoid, lateral deltoid, and posterior deltoid).	• One week after surgery if there are no surgical drains or after the surgical drains are removed. • At least twice a day until limb functions are returned to normal. • Whenever limb mobility is limited throughout the recovery.
Keep a healthy weight		
• Eat nutrition-balanced diet (ie, more vegetables and fruits as well as quality proteins). • Maintain portion-appropriate diet (feeling 75% full for each meal).	• Overweight or obesity is an important risk factor for lymph fluid accumulation. • Having extra weight makes it difficult for lymph flow and drain. This can lead to extra lymph fluid build-up. • There are numerous weight management programs available to assist with weight loss. • Although there are a lot of weight reduction programs, each person may respond differently to each program. • The core of the weight management is to eat a nutrition-balanced, portion-appropriate diet. It is also important to stay hydrated, exercise, and get adequate sleep.	• Each meal daily • It is important to talk to the nutritionist who can help to find a proper weight reduction program.
• Stay hydrated	• People may actually be thirsty, not hungry.	• Drink 6-8 glasses of water daily; in the morning, before and during meals, and throughout the day. • Avoid drinks with calories (eg, juices). • Drink green tea to boost metabolism.
• Large muscle exercises	• Daily large muscle exercises (eg, walking, running, swimming, yoga) help to burn more calories. • Daily large muscle exercises also promote lymph flow by creating muscle pumps.	• At least 30 minutes 3 times a week or daily

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Strategies and exercises	Rationales	Actions
• Get enough sleep	• Lack of sleep increases the production of the stress hormone cortisol, creates hunger, and leads to overeating. • Getting just 1 more hour of sleep per night reduces belly fat accumulation.	• At least 7-8 hours of sleep per night.

[a]TOLF: The-Optimal-Lymph-Flow.

The-Optimal-Lymph-Flow Intervention Group (n=60)

Patients assigned to the TOLF intervention group were granted the access to the web- and mobile-based TOLF platform to learn about the program and therapeutic lymphatic exercises during the first in-person research visit. Patients had the access to the website contents of Lymphedema, Diagnosis of Lymphedema, Lymphatic System, Self-care, Therapeutic Lymphatic Exercises, and Ask Experts. Patients also had the access to the 8 avatar videos with step-by-step instructions to perform lymphatic exercises to promote lymph flow and optimize shoulder and limb mobility. In addition, the patients were introduced to an app, and had the choice to use either the web-based program or the app for practicing lymphatic exercises. However, patients in the TOLF intervention group did not have the access to the section *Arm Precautions* because the participants in the TOLF intervention group received comparable information regarding self-care as in the *Arm Precautions* section but with particular emphasis on "what to do," rather than "what to avoid."

The Arm Precaution Control Group (n=60)

Patients assigned to the control AP group had access to the web- and mobile-based Arm Precaution program to learn about the program and therapeutic limb mobility exercises to promote limb mobility during the first in-person research visit. The AP program also focused on precautionary lifestyle behaviors, such as avoidance of repetitive limb movement, lifting weighted objects, needle punctures, blood draw, and the use of compression garments for air travel in the affected limb [20,21]. Patients had access to the following contents of the website: Lymphedema, Diagnosis of Lymphedema, Risk of Lymphedema, Lymphatic System, 5 avatar videos for Therapeutic Limb Mobility Exercises to promote limb mobility, and Arm Precautions.

Duration of Intervention

It took 30-45 minutes for patients to learn all the sections of the program and about 10 minutes to learn the TOLF lymphatic exercises for the intervention group through 8 avatar videos. It took about 5 minutes to perform a set of TOLF daily exercises each time. Participants in the AP control group had access to 5 limb mobility exercise avatar videos and it took 3 minutes to perform a set of limb mobility exercises each time. We encouraged patients to perform the assigned exercises at least twice a day during the 12-week study period.

Data Collection

Data Collection Procedures

Overview

Data were collected at baseline prior to the intervention, and at week 12 after the intervention. Data collection at each in-person time point took approximately 30 minutes. Within 1 week of enrollment for the clinical trial, patients had baseline assessment of pain and symptoms, limb volume difference, BMI, and QOL. The follow-up in-person assessment occurred at week 12 after the intervention.

Two In-Person Research Visits

Patients had 2 in-person research visits: (1) prior to the intervention: baseline assessment of pain and symptoms, limb volume difference, BMI, and QOL; and (b) week 12 postintervention assessment of pain and symptoms, limb volume difference, BMI, self-care behaviors, and QOL.

Two Online Assessments

Patients in the intervention and control groups received an email that provided a link to assess pain at weeks 4 and 8 after the intervention. Confidentiality of the patients was protected for the online assessment because patients used their study ID to access the online assessment.

Outcome Measures

Demographic and Medical Information

A structured tool was used to gather demographic and medical information and verified through reviewing participants' medical records [14,17,19]. The demographic and medical information included age, types of surgeries, lymph nodes procedure, radiation, chemotherapy, time since surgery, lymphedema diagnosis, and pain medications prior to and at week 12 after the intervention.

Primary and Secondary Outcome Measures

Primary measure focused on pain that was assessed prior to and at week 12 after the intervention during in-person visits as well as at weeks 4 and 8 postintervention online assessment. Secondary measures included symptoms, limb volume difference (measured using an infrared perometer), BMI, and QOL. Limb volume difference (measured using an infrared perometer) and BMI were measured prior to and at week 12 after the intervention during in-person visits. QOL was assessed prior to and at week 12 after the intervention during in-person visits as well as at weeks 4 and 8 after the online assessment.

Pain and Lymphedema Symptoms. The Lymphedema and Breast Cancer Symptom Experience Index(Part I) is a valid and reliable self-report tool to assess chronic pain, including aching,

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soreness, tenderness, and additional symptoms related to lymph fluid accumulation or lymphedema (ie, arm swelling, breast swelling, chest wall swelling, heaviness, firmness, tightness, stiffness, burning, stabbing numbness, tenderness, stiffness, redness, blistering, and tingling [pins and needles]) [14,17,19]. A response frame of last 3 months was used for all participants to ensure the chronicity of symptom presence during the first in-person visit prior to the intervention. A response frame of 7 days was used during the second in-person visit at week 12 after the intervention. Each item was rated on a Likert scale from 0 (no presence of a given symptom) to 4 (greatest severity of a given symptom). For this study, a complete pain reduction was defined when a patient's pain score was greater than 0 prior to the intervention and when the pain score was 0 at week 12 after the intervention.

Limb Volume Difference Measurement Using an Infrared Perometer

Perometry (350S; Juzo) was performed on each arm as it was held horizontally. The perometer maps a 3D graph of the affected and nonaffected extremities using numerous rectilinear light beams, and interfaces with a computer for data analysis and storage. A 3D limb image was generated and limb volume was calculated. This optoelectronic method has an SD of 8.9 mL (arm), <0.5% of limb volume with repeated measuring [17,19]. We used the following formula to calculate limb volume: limb volume difference percent = (affected limb volume – unaffected limb volume)/unaffected limb volume. An interlimb volume difference of >10% is a widely accepted diagnostic criterion for breast cancer–related lymphedema [10], yet it is known that a 5% difference in interlimb volume causes symptoms [24,25] and impairments in activities of daily living [27]. Therefore, we used the interlimb volume difference >5% as the threshold for minimal limb volume differences in this study.

General Bodily Pain and Quality of Life Related to Pain

The 6-item Pain Impact Questionnaire (PIQ-6), a reliable and valid 6-question health survey, was used to measure the impact of pain on an individual's functional health and well-being. The PIQ-6 measures the severity of general bodily pain and its impact on work and leisure activities, as well as on emotional well-being within a variety of diseases and general populations. High PIQ-6 *t*-scores indicate greater pain impact/worse health [28].

Height, Body Weight, and BMI

Height was measured to the nearest 0.1 cm with a portable stadiometer (Scale-Tronix 5002 Stand on Scale; Scale-Tronix Company) without shoes [29]. An electrical device (InBody 520, Biospace Co., Ltd) was used to measure the participants' body weight, and BMI was calculated using the formula: weight (kg)/height (m^2) [29].

Practice of Self-care Behaviors

The Risk Reduction Behavior Checklist, a structured self-report checklist, was used to quantitatively assess patients' self-report of adherence to the assigned interventions at the study endpoint of 12 weeks after the intervention [17,19]. The checklist included a list of self-care behaviors that promote lymph flow (eg, muscle tightening–deep breathing, muscle tightening–pumping, limb mobility exercises).

Statistical Analysis

Primary Endpoint

The primary endpoint for the study was a complete pain reduction or reduced pain severity reported by the participants at week 12 after the intervention.

Sample Size and Power Calculations

The target sample size was 120 participants to account for a potential attrition of 20%, which has been observed in previous studies on breast cancer survivors [10]. This allowed to yield an adequate analytic sample size even with 20% attrition based on a 2-sample 2-sided *t* test with α=.05 and power of 90% to detect a group difference of 0.7 SDs in pain severity reported by the participants at week 12 after the intervention. The projected sample size of 96 would also provide sufficient statistical power for mixed regression models and for linear mixed models of continuous outcomes (eg, QOL).

Data Analysis

Data downloading and entry were performed independently by 2 researchers who were not involved in data collection and had no conflicts of interest. Moreover, the data analysis was independently assessed by 2 experienced statisticians (MM and LF) who were not involved in the data collection. Data were analyzed using R version 3.6.2 (R Foundation for Statistical Computing). Descriptive statistics were performed for baseline demographic and clinical characteristics using parametric (eg, independent samples *t* test) and nonparametric tests (eg, chi-square test) as appropriate. All the tests were 2 tailed. Descriptive statistics were also performed to summarize the distributions for primary and secondary outcome variables.

As planned [30], Fisher exact tests were used to test the primary hypothesis that more patients who received the TOLF intervention would report a complete reduction of chronic pain, aching, soreness, tenderness, and general bodily pain compared with patients who received the AP intervention at week 12 after the intervention. Wilcoxon rank sum tests were performed to test the hypothesis that patients who received the TOLF intervention would report less severe chronic pain, aching, soreness, tenderness, and general bodily pain at week 12 after the intervention compared with patients who received the AP control intervention. The proportion of patients reporting complete pain reduction was compared between the TOLF intervention and AP control groups prior to the intervention and at week 12 after the intervention using Fisher exact tests.

As planned [30], additional mixed effects models were conducted to test for between-group differences in change of pain over the study period. Generalized linear mixed effects models (cumulative logit mixed models) were used to analyze ordinal outcomes (eg, ratings pain, aching, soreness, tenderness, general bodily pain) and generalized linear mixed models (binomial mixed effects models with a logit link) were used to analyze binary outcomes (presence of pain, aching, soreness, tenderness, general bodily pain). These models incorporated fixed effects for time, treatment group, and a time × group

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interaction term, as well as a random intercept to account for repeated within-person observations. The models were estimated using maximum likelihood with adaptive Gaussian quadrature approximation methods.

To test the secondary hypothesis that patients who received the TOLF intervention would report fewer lymphedema symptoms, minimal limb volume differences, and better BMI and QOL compared with patients who received the AP control intervention, independent sample *t* tests for numeric continuous variables and chi-square or Fisher exact tests for nominal variables were used to assess the changes between the group differences in secondary outcomes between the TOLF intervention and AP control groups at week 12 after the intervention. We supplemented each of these comparisons with between-group tests prior to the intervention for reference.

Method of Handling Missing Data and Nonadherence to Protocol

There was no case of nonadherence to study protocol. No participants had a missing data >20%. Data were missing from the 6 patients due to attrition. Other participants have intermittent missing data throughout the study due to nonresponse. All missing data were not systematic but missing at random. The primary objective of this RCT required nonparametric tests, precluding the use of Rubin's rules for multiple imputation and intent-to-treat analysis [31]. These results were all based on complete cases, and inferences represent effects of treatment on the treated. In addition, linear mixed effects models with maximum likelihood estimation were used to address between-group differences in pain, aching, soreness, and tenderness during the intervention. These analyses were in accordance with intent-to-treat principles [31].

Results

Participant Characteristics at Baseline

Among the 120 enrolled patients, 114 participants completed the study, including 1 case of screen failure. This patient in the control group was deemed ineligible but completed the study because she was diagnosed with other cancer before the end of the study (0.8% [1/114] screen failure). At week 12 after the intervention, 5 participants in the intervention group and 1 participant in the control group did not complete the study (5% [6/120] attrition rate). There were no statistical differences in demographic and treatment characteristics between patients that completed the study and the 6 patients who did not. No statistical differences were also found between participants in the TOLF intervention and AP control groups in terms of demographic and treatment characteristics except that participants in the TOLF intervention group had higher weight compared with those in the AP control group at baseline prior to the intervention. As a result, the randomization scheme based on the presence of pain, aching, soreness, or tenderness created 2 relatively similar patient profiles (Table 2).

As shown in Table 2, at baseline, the participants were women with a mean age of 56.7 years (SD 10.6; range 54.7-58.6). More than 70% (88/120, 73.3%) had a bachelor or graduate/professional degree, 50.8% (61/120) were married, and 65.8% (79/120) were employed. Of the 120 patients, 70% (84/120) had lumpectomy while 30% (36/120) had mastectomy, 59.2% (71/120) had chemotherapy, and 77.5% (93/120) had radiation therapy. While 32.5% (39/120) of the patients underwent axillary lymph nodes dissection, 59.2% (71/120) had sentinel lymph nodes biopsy alone. The mean lymph nodes removed was 7.3 (SD 7.7; range 5.9-9.0). Only 15.8% (19/120) of the participants had been diagnosed with and treated for lymphedema. There was a 13% reduction (from 40% [24/60] to 27% [16/60]) in proportions of patients who took pain medications in the TOLF intervention compared with a 5% increase (from 40% [24/60] to 45% [27/60]) in the control group at week 12 after the intervention.

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Table 2. Demographic and clinical characteristics of participants at baseline (N=120).

Characteristics	Total (N=120), mean (SD), range	Arm precaution (n=60)	The-Optimal-Lymph-Flow (n=60)	Statistics (*df*)	*P* value[a]
Age (years), mean (SD), range	56.7 (10.6), 54.7-58.6	56.8 (11.0), 53.9-59.6	56.6 (10.3), 53.9-59.2	$t_{116.69}$=–0.11	.91
Body weight (lb), mean (SD), range	163.58 (38.2), 156.6-170.5	156.2 (39.0), 146.1-166.2	171.1 (36.2), 161.7-180.5	$t_{116.59}$=2.17	.03
Number of lymph nodes removed, mean (SD), range	7.3 (7.7), 5.9-9.0	7.4 (8.7), 5.1-9.7	7.2 (6.5), 5.5-9.0	$t_{103.46}$=–0.11	.91
Time since breast cancer diagnosis (years) to study enrollment, mean (SD), range	2.8 (1.2), 2.6-3.0	2.7 (1.2), 2.4-3.0	2.9 (1.2), 2.5-3.2	$t_{114.65}$=0.62	.54
Level of education, n (%)				Fisher exact test (5)	.79
High school or below	26 (21.7)	12 (20.0)	14 (23.3)		
Associate's degree	6 (5.0)	3 (5.0)	3 (5.0)		
Bachelor's degree	47 (39.2)	23 (38.3)	24 (40.0)		
Master's degree	29 (24.2)	17 (28.3)	12 (20.0)		
Doctoral degree	7 (5.8)	2 (3.3)	5 (8.3)		
Professional degree	5 (4.2)	3 (5.0)	2 (3.3)		
Marital status, n (%)				Fisher exact test (4)	.16
Married	61 (50.8)	26 (43.3)	35 (58.3)		
Divorced/separated	16 (13.3)	11 (18.3)	5 (8.3)		
Widowed	7 (5.8)	3 (5.0)	4 (6.7)		
Partnered	9 (7.5)	3 (5.0)	6 (10.0)		
Single or never partnered	27 (22.5)	17 (28.3)	10 (16.7)		
Ethnicity, n (%)				Fisher exact test (4)	.98
Asian	10 (8.3)	5 (8.3)	5 (8.3)		
African American or Black	22 (18.3)	12 (20.0)	10 (16.7)		
White	72 (60.0)	35 (58.3)	37 (61.7)		
Hispanic/Latino	11 (9.2)	5 (8.3)	6 (10.0)		
More than 1 race	5 (4.2)	3 (5.0)	2 (3.3)		
Employment status, n (%)				χ_1^2=0.07	.80
Unemployed	41 (34.2)	19 (32)	22 (36.7)		
Employed	79 (65.8)	41 (68)	38 (63.3)		
Mastectomy				χ_1^2=0.07	.84
Yes	36 (30.0)	19 (31.7)	17 (28.3)		
No	84 (70.0)	41 (68.3)	43 (71.7)		
Lumpectomy					>.99
Yes	60 (50.0)	30 (50.0)	30 (50.0)		
No	60 (50.0)	30 (50.0)	30 (50.0)		
Being diagnosed with and treated for lymphedema, n (%)				Fisher exact test (1)	.32
Yes	19 (15.8)	12 (20.0)	7 (11.7)		
No	98 (81.7)	47 (78.3)	51 (85.0)		

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Characteristics	Total (N=120), mean (SD), range	Arm precaution (n=60)	The-Optimal-Lymph-Flow (n=60)	Statistics (*df*)	*P* value[a]
Radiotherapy, n (%)				χ_1^2=1.13	.29
Yes	93 (77.5)	44 (73.3)	50 (83.3)		
No	27 (22.5)	16 (26.7)	10 (16.7)		
Chemotherapy, n (%)				χ_1^2=0.412	.52
Yes	71 (59.2)	33 (55.0)	38 (63.3)		
No	49 (40.8)	27 (45.0)	22 (36.7)		
Axillary lymph nodes dissection, n (%)				χ_1^2<0.001	>.99
Yes	39 (32.5)	20 (33.3)	19 (31.7)		
No	27 (22.5)	14 (23.3)	13 (21.7)		
Sentinel lymph nodes biopsy alone, n (%)				Fisher exact test (1)	.71
Yes	71 (59.2)	40 (66.7)	41 (68.3)		
No	49 (40.8)	20 (33.3)	19 (31.7)		
Taking pain medications at baseline, n (%)				χ_1^2<0.001	>.99
Yes	48 (40.0)	24 (40.0)	24 (40.0)		
No	72 (60.0)	36 (60.0)	36 (60.0)		
Taking pain medications at week 12 after the intervention, n (%)				χ_1^2=0.344	.56
Yes	43 (35.8)	27 (45.0)	16 (26.7)		
No	76 (63.3)	33 (55.0)	44 (73.3)		

[a]*P* values were derived from independent samples *t* tests for numeric outcomes. For categorical outcomes, *P* values correspond to chi-square tests of independence unless any cell sizes are <10, in which case a Fisher exact test was performed.

Complete Reduction of Pain, Aching, Soreness, Tenderness, and General Bodily Pain

As shown in Table 3, at baseline prior to the intervention, there were no significant differences between the TOLF intervention and AP control groups in terms of proportions of patients who reported chronic pain (*P*>.99), aching (*P*=.42), soreness (*P*=.12), tenderness (*P*=.28), and general bodily pain (*P*>.37). At the study endpoint of week 12, significantly fewer patients in the TOLF intervention group compared with the AP control group reported chronic pain (45% [27/60] vs 70% [42/60]; odds ratio [OR] 0.39, 95% CI 0.17-0.90; *P*=.02). No significant differences were found between the TOLF and AP groups in terms of proportion of patients who reported aching (*P*=.05), soreness (*P*=.12), or tenderness (*P*=.25) as well as general bodily pain (*P*=.28).

As presented in Table 4, Fisher exact tests demonstrated that patients who received the TOLF intervention were more likely to experience a complete reduction in chronic pain (50% [23/46] vs 22% [11/51]); OR 3.56, 95% CI 1.39-9.76; *P*=.005) and soreness (43% [21/49] vs 22% [11/51]; OR 2.60, 95% CI 1.03-6.81; *P*=.03) compared with patients who received the AP control at week 12 after the intervention. There were no significant differences in complete reduction of aching (*P*=.12), tenderness (*P*=.65), and general bodily pain (*P*=.16) between the TOLF and AP groups.

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Table 3. Proportion of patients that reported chronic pain, soreness, aching, or tenderness, and general bodily pain at baseline prior to the intervention and at study endpoint of week 12 after the intervention.

Outcome variables	Arm precaution (n=60)		The-Optimal-Lymph-Flow (n=60)		Fisher exact test of independence[a]	
	No pain, n (%)	Pain, n (%)	No pain, n (%)	Pain, n (%)	Odds ratio (95% CI)	*P* value
Baseline prior to the intervention						
Chronic pain	8 (13)	52 (87)	9 (15)	51 (85)	0.87 (0.27-2.77)	>.99
Soreness	5 (8)	55 (92)	6 (10)	54 (90)	0.82 (0.19-3.44)	>.99
Aching	6 (10)	53 (88)	10 (17)	49 (82)	0.56 (0.15-1.84)	.42
Tenderness	8 (13)	52 (87)	12 (20)	47 (78)	0.61 (0.20-1.77)	.34
General bodily pain	3 (5)	57 (95)	1 (2)	59 (98)	1.04[b] (0.97-1.11)	.37
Week 12 after the intervention						
Chronic pain	17 (28)	42 (70)	28 (47)	27 (45)	0.39 (0.17-0.90)	.02[c]
Soreness	17 (28)	42 (70)	24 (40)	31 (52)	0.53 (0.22-1.22)	.12
Aching	17 (28)	42 (70)	26 (43)	28 (47)	0.44 (0.19-1.01)	.05
Tenderness	19 (32)	40 (67)	24 (40)	31 (52)	0.62 (0.27-1.41)	.25
General bodily pain	11 (18)	48 (80)	15 (25)	40 (67)	0.89[b] (0.73-1.09)	.28

[a]Degrees of freedom (*df*)=1 for all the tests.

[b]Because general bodily pain is a very likely outcome prior to the intervention, we report risk ratio rather than odds ratio for this outcome at both time points. The 95% CI corresponds to the risk ratio, and *P* values are obtained using Monte Carlo simulation.

[c]Statistical significance.

Table 4. Proportions of patients with complete pain reduction between the intervention and control groups ("Yes"= complete pain reduction) using Fisher exact tests.

Complete pain reduction	Arm precaution		The-Optimal-Lymph-Flow		Test of group differences		
	No, n (%)[a]	Yes, n (%)	No, n (%)	Yes, n (%)	OR[b] (95% CI)	*P* value	NNT[c]
Chronic pain	40 (78)	11 (22)	23 (50)	23 (50)	3.56 (1.39-9.76)	.005[d]	3.6
Tenderness	38 (75)	13 (25)	29 (69)	13 (31)	1.31 (0.48-3.56)	.65	16.7
Soreness	42 (78)	11 (22)	28 (57)	21 (43)	2.60 (1.03-6.81)	.03[e]	4.8
Aching	40 (77)	12 (23)	26 (60)	17 (40)	2.16 (0.82-5.86)	.12	5.9
General bodily pain	48 (86)	8 (14)	40 (74)	14 (26)	2.09 (0.73-6.36)	.16	8.3

[a]Percentage is based on numbers of patients who reported chronic pain, tenderness, soreness, aching, and general bodily pain at baseline. That is, denominator for each symptom is different. For example, for chronic pain for the arm precaution group N is 51. There were 51 patients in the arm precaution group who reported nonzero chronic pain at visit 1.

[b]OR: odds ratio, a measure of effect size. Recommended interpretation: 1.5=small, 2=medium, 3=large. Degrees of freedom (*df*)=1 for all the tests.

[c]NNT: number needed to treat, that is, the number of patients who would need to participate in the TOLF intervention (instead of the AP control) for 1 additional patient to experience a complete pain reduction.

[d]Significant at the *P*<.01 level.

[e]Significant at the *P*<.05 level.

Severity of Chronic Pain, Aching, Soreness, Tenderness, and General Bodily Pain

At baseline, there were no significant differences in terms of median severity of chronic pain (*P*=.08), aching (*P*=.05), soreness (*P*=.07), tenderness (*P*=.13), or general bodily pain (*P*=.56) between the TOLF intervention and AP control groups (Table 5). At week 12 after the intervention, the TOLF group had significantly lower median severity scores for chronic pain (Med_{TOLF}=0, IQR=0-1 vs Med_{AP}=1, IQR 0-2; *P*=.02) and general bodily pain (Med_{TOLF}=1, IQR 0-1.5 vs Med_{AP}=1, IQR=1-3; *P*=.04).

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Table 5. Severity of chronic pain, soreness, aching, or tenderness, and general bodily pain as well as quality of life (PIQ-6) at baseline prior to the intervention and study endpoint of week 12 after the intervention.

Outcome variables	Arm precaution (n=60), median (IQR)	The-Optimal-Lymph-Flow (n=60), median (IQR)	Independent samples test for between-group differences		
			Wilcoxon r^a (95% CI)	W-score	*P* value
Baseline prior to the intervention					
Chronic pain	2 (1-3)	1 (1-2)	0.161 (–0.029 to 0.334)	2125	.08
Soreness	2 (1-3)	2 (1-2)	0.165 (–0.008 to 0.346)	2133	.07
Aching	2 (1-3)	2 (1-2)	0.177 (–0.008 to 0.346)	2089	.05
Tenderness	2 (1-3)	2 (1-3)	0.139 (–0.035 to 0.328)	2048	.13
General bodily pain	2 (1.75-3)	2 (2-3)	0.054 (–0.138 to 0.233)	1908.5	.56
Quality of life[b] by PIQ-6[c]	56.1 (9.3)	54.1 (7.5)	0.234 (–0.125 to 0.601)	1.30 (112.9)	.20
Week 12 after the intervention					
Chronic pain	1 (0-2)	0 (0-1)	0.206 (0.030 to 0.378)	2001	.02[d]
Soreness	1 (0-2)	1 (0-2)	0.117 (–0.071 to 0.292)	1837	.20
Aching	1 (0-2)	1 (0-1.75)	0.160 (–0.032 to 0.335)	181.5	.08
Tenderness	1 (0-2)	1 (0-2)	0.055 (–0.121 to 0.237)	1723.5	.55
General bodily pain	1 (1-3)	1 (0-1.5)	0.188 (0.016 to 0.355)	1968	.04[d]
Quality of life[b] by PIQ-6	50.7 (8.1)	48.4 (7.9)	0.290 (–0.088 to 0.669)	1.53 (108.5)	.13

[a]Wilcoxon *r*: Measure of effect size. Recommended interpretation: 0.1=small, 0.3=medium, 0.5=large.

[b]For quality of life, data in columns 2-6 are presented as mean (SD), mean (SD), Cohen *d* (95% CI), t-score (*df*), and *P* value. Cohen *d* is a measure of effect size. Recommended interpretation: 0.2=small, 0.5=medium, 0.8=large.

[c]PIQ-6: 6-item Pain Impact Questionnaire.

[d]Significant at the *P*<.05 level.

Changes of Pain, Aching, Soreness, Tenderness, and General Bodily Pain

Cumulative link mixed effects models were used to predict the ordinal pain outcomes (pain, soreness, aching, tenderness) across the 4 measurement time points and to determine group differences in the changes during the study time. As shown in Multimedia Appendix 2, there was a significant decrease in severity of chronic pain, aching, soreness, and tenderness for both the TOLF intervention and AP control groups across the 4 time points (baseline, week 4, 8, and 12 after the intervention). There was no significant time × group interaction effect for chronic pain (*P*=.14), aching (*P*=.23), soreness (*P*=.22), and tenderness (*P*=.18).

Binomial mixed effects models were used to assess group differences in the prevalence of chronic pain, aching, soreness, and tenderness across the study time points. Model results (Multimedia Appendix 3) indicate that patients were less likely to experience chronic pain, tenderness, soreness, and aching throughout the course of the study. This effect was consistent for both the TOLF intervention and AP control groups. There were no group differences.

A cumulative link mixed effects model was also used to predict severity of general bodily pain across the 4 time points and to determine whether the 2 groups differed in how pain scores vary across the 4 study time points. As Multimedia Appendix 4 shows, there was a significant decrease in general bodily pain across the 4 time points. This effect was consistent for both the TOLF intervention and AP control groups. There was no significant time × group interaction (*P*=.22). Results of the binomial mixed effects model to assess group differences in the prevalence of general bodily pain across the study period are shown in Multimedia Appendix 4. Patients were less likely to experience general bodily pain throughout the course of the study. This effect was similar between the TOLF intervention and AP control groups.

Quality of Life Related to Pain

At baseline prior to the intervention, there was no significant difference ($t_{112.9}$=1.30, *P*=.20) in mean QOL by PIQ-6 scores between the TOLF intervention (54.1 [SD 7.5]) and AP control (56.1 [SD 9.3]). At the study endpoint of week 12, there was no significant difference in mean QOL by PIQ-6 scores ($t_{108.5}$=1.53, *P*=.13) between the TOLF intervention (48.4 [SD 7.9]) and the AP control (50.7 [SD 8.1]). As more improvement in the PIQ-6 scores was found in the TOLF intervention group at week 12 after the intervention, we conducted a subsequent linear mixed effects model predicting PIQ-6 scores across the 4 study time points and confirmed that PIQ-6 scores were significantly improved during the study (*B*=–1.73, 95% CI –2.33 to –1.13, *P*<.001) in the TOLF and AP groups, and that changes

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in PIQ-6 were not statistically different between the TOLF intervention and AP control groups (*b*=0.07, 95% CI –0.80 to 0.93; *P*=.88; Multimedia Appendix 5).

Lymphedema Symptoms, Limb Volume Differences, and BMI

Table 6 presents the occurrence of the 23 lymphedema symptoms at baseline prior to and after the intervention. Compared with the AP control group, at week 12 after the intervention, significantly fewer patients in the TOLF intervention group reported arm/hand swelling (*P*=.04), heaviness (*P*=.03), redness (*P*=.03), and limited movement in shoulder (*P*=.02) and arm (*P*=.03). As shown in Table 7, there were no significant differences at week 12 after the intervention between the TOLF intervention and AP control groups in terms of mean numbers of lymphedema symptom reported, ≥5% limb volume differences, and BMI. There was a 12% reduction (from 27% [16/60] to 15% [9/60]) in the proportion of patients with ≥5% limb volume differences from baseline to postintervention in the TOLF group, while there was a 5% increase (from 40% [24/60] to 45% [27/60]) in the proportion of patients with ≥5% limb volume differences from baseline to postintervention in the AP group.

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Table 6. Lymphedema symptom occurrence at baseline and after the intervention.

Lymphedema symptoms	Arm precaution (n=60), n (%)	The-Optimal-Lymph-Flow (n=60), n (%)	*P* value[a]
Arm/hand swelling			
Baseline	34 (57)	30 (50)	.46
Week 12 after the intervention	28 (47)	17 (28)	.04[b]
Breast swelling			
Baseline	23 (38)	30 (50)	.57
Week 12 after the intervention	14 (23)	13 (22)	.83
Chest wall swelling			
Baseline	7 (12)	10 (17)	.36
Week 12 after the intervention	8 (13)	8 (13)	>.99
Firmness in the affected limb			
Baseline	19 (32)	16 (27)	.57
Week 12 after the intervention	21 (35)	16 (27)	.55
Tightness in the affected limb			
Baseline	29 (48)	30 (50)	.49
Week 12 after the intervention	24 (40)	22 (37)	.76
Heaviness in the affected limb			
Baseline	26 (43)	20 (33)	.40
Week 12 after the intervention	26 (43)	15 (25)	.03[b]
Toughness of thickness of skin in the affected limb			
Baseline	14 (23)	9 (15)	.35
Week 12 after the intervention	10 (17)	8 (13)	.60
Stiffness in the affected limb			
Baseline	25 (42)	22 (37)	.61
Week 12 after the intervention	29 (48)	18 (30)	.10
Hotness/increased temperature in the affected limb			
Baseline	13 (22)	9 (15)	.47
Week 12 after the intervention	14 (23)	7 (12)	.13
Redness in the affected limb			
Baseline	7 (12)	4 (7)	.38
Week 12 after the intervention	8 (13)	1 (2)	.03[b]
Blistering in the affected limb			
Baseline	3 (5)	1 (2)	.62
Week 12 after the intervention	0 (0)	0 (0)	N/A[c]
Numbness in the affected limb			
Baseline	20 (33)	31 (52)	.01
Week 12 after the intervention	18 (30)	19 (32)	.96
Burning in the affected limb			
Baseline	4 (7)	12 (20)	.03
Week 12 after the intervention	3 (5)	9 (15)	.10
Stabbing in the affected limb			
Baseline	13 (22)	10 (17)	.65

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Lymphedema symptoms	Arm precaution (n=60), n (%)	The-Optimal-Lymph-Flow (n=60), n (%)	*P* value[a]
Week 12 after the intervention	13 (22)	8 (13)	.19
Tingling (pins and needles) in the affected limb			
Baseline	25 (42)	32 (53)	.40
Week 12 after the intervention	20 (33)	23 (38)	.74
Fatigue in the affected limb			
Baseline	23 (38)	31 (52)	.29
Week 12 after the intervention	21 (35)	29 (48)	.15
Weakness in the affected limb			
Baseline	35 (58)	22 (37)	.05
Week 12 after the intervention	34 (57)	21 (35)	.02[b]
Seroma (pocket or fluid developed)			
Baseline	10 (17)	8 (13)	.76
Week 12 after the intervention	9 (15)	3 (5)	.08
Limited movement in shoulder			
Baseline	23 (38)	22 (37)	.84
Week 12 after the intervention	28 (47)	15 (25)	.02[b]
Limited movement in elbow			
Baseline	9 (15)	6 (10)	.48
Week 12 after the intervention	11 (18)	5 (8)	.15
Limited movement in wrist			
Baseline	15 (25)	11 (18)	.53
Week 12 after the intervention	15 (25)	7 (12)	.07
Limited movement in fingers			
Baseline	21 (35)	14 (23)	.27
Week 12 after the intervention	17 (28)	9 (15)	.08
Limited movement in arm			
Baseline	26 (43)	27 (45)	.46
Week 12 after the intervention	27 (45)	15 (25)	.03[b]

[a]Chi-square tests of independence unless any cell sizes are <10, in which case a Fisher exact test was performed.

[b]Significant at $P<.05$.

[c]N/A: not applicable.

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Table 7. Outcomes of lymphedema symptoms, limb volume differences, and BMI at baseline and after the intervention.[a]

Secondary outcome variables	The-Optimal-Lymph-Flow (n=60)	Arm precaution (n=60)	Statistics	*P* value[b]
Number of lymphedema symptoms				
Baseline	9.2 (5.4), 7.8-10.6	10.6 (4.9), 9.3-11.8	$t_{116.96}$=–1.48	.14
Week 12 after the intervention	6.1 (5.1), 4.6-7.4	7.6 (5.2), 6.2-8.9	$t_{111.61}$=–1.62	.11
Mean BMI				
Baseline prior to the intervention	29.2 (6.0), 27.6-30.8	27.1 (6.4), 25.4-28.8	$t_{115.86}$=1.86	.07
Week 12 after the intervention	29.3 (6.3), 27.6-31.1	27.4 (6.5), 5.7-29.1	$t_{107.49}$=1.58	.12
≥5% Limb volume differences[c]			χ_1^2=0.230	
Baseline				.63
Yes	16 (27)	13 (22)		
No	44 (73)	47 (78)		
Week 12 after the intervention[d]				.48
Yes	9 (15)	16 (27)		
No	51 (85)	44 (73)		

[a]Data are presented as mean (SD), range or n (%).

[b]*P* values are derived from independent samples *t* tests for numeric outcomes. For categorical outcomes, *P* values correspond to chi-square tests of independence unless any cell sizes are <10, in which case a Fisher exact test was performed.

[c]Limb volume difference percent = (affected limb volume – unaffected limb volume)/unaffected limb volume.

[d]Fisher exact test (*df*=1) was applied.

Self-report of Adherence

Participants reported no adverse events of performing the TOLF lymphatic exercises and limb mobility exercises. In terms of self-reported adherence to the assigned interventions, 87% (52/60) of participants reported performing the TOLF lymphatic exercises twice a day as prescribed, while 83% (50/60) of participants reported performing limb mobility exercises twice a day as prescribed.

Discussion

Preliminary Findings

The therapeutic lymphatic and limb mobility exercise intervention is an essential component of the TOLF self-care pain management program [19,23]. The efficacy of the TOLF intervention relies on skill-based training in teaching patients to correctly perform the set of therapeutic exercises. Prior research identified that ambiguous and inadequate information was a barrier to initiate and maintain exercise for breast cancer survivors [32]. Clear information about how exercise should be done and how often it should be done is essential for patients to initiate and adhere to the prescribed therapeutic exercise regimen [33,34]. Extending prior research findings [23,35], this RCT provided additional evidence that the web- and mobile-based TOLF system is feasible and efficacious in training patients to perform lymphatic and limb mobility exercises via avatar videos with step-by-step instructions.

A recent a single-arm feasibility clinical trial with a pre- and posttest design to assess the effects of the TOLF therapeutic lymphatic exercise intervention demonstrated that a single session of a Kinect-enhanced TOLF intervention immediately reduces pain, swelling, and lymphedema symptoms in breast cancer survivors [35]. This current RCT was the first to evaluate the effectiveness of the web- and mobile-based TOLF system for managing chronic pain and lymphedema symptoms by comparing 2 parallel interventions. Results of this RCT demonstrated that the TOLF intervention to promote lymph flow led to more complete pain reductions and pain severity reductions at week 12 after the intervention compared with the AP control to improve limb mobility. The TOLF intervention achieved a large effect for complete reduction in pain (OR 3.56, 95% CI 1.39-9.76; *P*=.005) and a medium effect for complete reduction in soreness (OR 2.60, 95% CI 1.03-6.81; *P*=.03).

Current pain management relies heavily on pharmacological agents, such as opioids and nonsteroidal anti-inflammatory drugs [30,31], which were also the major pain medications that our participants took. It is important to note that a 13% reduction (from 40% [24/60] to 27% [16/60]) was observed in proportions of patients who took pain medications at week 12 after the intervention in the TOLF intervention, while a 5% increase in the AP control (from 40% [24/60] to 45% [27/60]) was noted. This result is promising due to concerns of poor efficacy, abuse, and adverse effects of opioids and nonsteroidal anti-inflammatory drugs [36,37]. Results of this RCT extend findings of prior single-arm trials [19,35] and suggest that the TOLF intervention is superior to the AP control in pain management.

Managing pain and lymphedema symptoms is critical to reduce the risk of lymphedema. Breast cancer survivors who report pain on the affected ipsilateral upper limb or body are nearly

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twice as likely to develop lymphedema [9]. For breast cancer survivors without a diagnosis of lymphedema, the experience of pain and lymphedema symptoms is a cardinal sign of subclinical lymphedema [38,39]. In this RCT, only 15.8% (19/120) of participants were diagnosed with or treated for lymphedema, yet all the participants without a diagnosis of or treated for lymphedema reported chronic pain and lymphedema symptoms at baseline prior to the intervention. Symptoms of arm/hand swelling, heaviness, redness, and limited movement in shoulder are hallmarks of fluid accumulation [28]. In this RCT, significantly fewer patients in the TOLF intervention group reported arm/hand swelling, heaviness, redness, and limited movement in shoulder and arm at the end of the trial. Extending findings of prior single-arm clinical trials [35,39], this RCT suggests that the TOLF intervention may be more effective than AP to effectively manage pain and lymphedema symptoms.

The TOLF lymphatic exercises were designed to decrease lymph fluid levels. In a previous study [19], 97% of the 134 patients who received the face-to-face TOLF intervention maintained or decreased their preoperative limb volumes assessed using an infrared perometer at 12 months after surgery. It is important to note that a 12% reduction (from 27% [16/60] to 15% [9/60]) in proportions of patients with ≥5% limb volume differences was observed in the TOLF group, whereas a 5% increase in the AP group (from 40% [24/60] to 45% [27/60]) was observed. This finding suggests that the TOLF intervention may be more effective in reducing limb volume than the AP control. In a recent study, significant reductions were found in lymph fluid levels assessed using bioimpedance immediately after a single training session of a Kinect-enhanced TOLF intervention [35]. More importantly, greater reductions in lymph fluid levels were found in patients with abnormal lymph fluid levels. The use of bioimpedance for assessing lymph fluid level may be a more sensitive measure than limb volume measurement using a perometer and should be applied in future studies.

Strengths and Limitations

The strengths of the RCT are a safe novel digital intervention targeting the lymphatic system for chronic pain, 5% (6/120) attrition, rigorous study design with a larger sample size over 100 patients, and the consecutively identified participants with chronic pain. The use of technologically driven digital therapy not only enhanced the fidelity and transparency of the intervention delivery but also the reproducibility of the intervention, which may enhance the generalizability and dissemination of the intervention. The technologically driven delivery model enhanced the patients' ability to learn to perform the assigned exercise therapy given that they were able to review the assigned exercise therapy on their own schedule and pace virtually anytime and anywhere. Another strength was the daily 5-minute routine of TOLF lymphatic exercises, which was easy for patients to establish in their own routine.

There were fewer limitations of this RCT. In our study, 87% [52/60] patients reported performing the TOLF lymphatic exercises twice a day as prescribed and 83% [50/60] reported performing AP limb mobility exercises. Lack of real-time monitoring limited the study's ability to explore dose sensitivity for pain. Future study may use wearable devices to monitor patients' adherence. Accumulation of lymph fluid in the affected area or limb leads to chronic inflammation resulting in pain for breast cancer survivors [28,29]. Pain following breast cancer treatment is significantly associated with the inflammatory cytokine gene *IL13* and lymphatic gene *VEGFC* [24,25]. Future research should investigate the genetic impact on the TOLF intervention as well as the efficacy of TOLF on the genetic expression of biomarkers.

Conclusions

The results of this RCT showed significant benefits of the TOLF intervention for chronic pain, soreness, general bodily pain, and specific lymphedema symptoms (ie, arm/hand swelling, heaviness, limited movement in shoulder and arm) among breast cancer survivors in comparison with the AP control. The TOLF intervention resulted in a 13% reduction (from 40% [24/60] to 27% [16/60]) in proportions of patients who took pain medications compared with the AP control, which had a 5% increase (from 40% [24/60] to 45% [27/60]). In addition, a 12% reduction (from 27% [16/60] to 15% [9/60]) in proportions of patients with ≥5% limb volume differences was observed in the TOLF group and a 5% increase in proportions of patients in the AP group (from 40% [24/60] to 45% [27/60]). These findings suggest that the TOLF intervention should be a better choice for pain management and limb volume reduction in comparison to the AP control. The TOLF intervention is safe, efficacious, and affordable as a replacement or complement therapy for chronic pain management for millions of breast cancer survivors. The low-cost, detailed description of interventions, and technologically driven delivery model of the TOLF make it relatively easy to implement TOLF in clinical practice or at home.

Acknowledgments

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Conflicts of Interest

None declared.

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Multimedia Appendix 1
CONSORT-EHEALTH checklist.
[PDF File (Adobe PDF File), 558 KB - cancer_v8i1e29485_app1.pdf]

Multimedia Appendix 2
Results of the cumulative link mixed effects models for ordinal pain outcomes. Time is centered at baseline prior to intervention=0.
[DOCX File , 17 KB - cancer_v8i1e29485_app2.docx]

Multimedia Appendix 3
Results of the binomial mixed effects models with logit link for binary pain outcomes (0=no, 1=yes). Time is centered at baseline prior to intervention=0.
[DOCX File , 15 KB - cancer_v8i1e29485_app3.docx]

Multimedia Appendix 4
Results for the cumulative link mixed effects model for ordinal general bodily pain (left) and the binomial mixed effects model for prevalence (0=no, 1=yes) of general bodily pain (right). Time is centered at baseline prior to intervention=0.
[DOCX File , 15 KB - cancer_v8i1e29485_app4.docx]

Multimedia Appendix 5
Results from a linear mixed effects model predicting PIQ-6 (Quality of Life) scores. Time is centered at baseline prior to the intervention=0.
[DOCX File , 14 KB - cancer_v8i1e29485_app5.docx]

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Abbreviations

AP: arm precaution
mHealth: mobile health
OR: odds ratio
PIQ: Pain Impact Questionnaire
QOL: quality of life
RCT: randomized clinical trial
TOLF: The-Optimal-Lymph-Flow

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Original Paper

An mHealth App to Support Caregivers in the Medical Management of Their Child With Cancer: Co-design and User Testing Study

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Abstract

Background: Caregivers face new challenges and tasks when their child is diagnosed with cancer, which can be overwhelming. Mobile technology has the capacity to provide immediate support at their fingertips to aid in tracking symptoms, managing medication, and planning for emergencies.

Objective: The objective of this study is to engage directly with end users and proxies to co-design and create a mobile technology app to support caregivers in the medical management of their child with cancer.

Methods: We engaged directly with caregivers of children with cancer and pediatric oncology nurse coordinators (proxy end users) to co-design and create the prototype of the Cope 360 mobile health app. Alpha testing was accomplished by walking the users through a series of predetermined tasks that encompassed all aspects of the app including tracking symptoms, managing medications, and planning or practicing for a medical emergency that required seeking care in the emergency department. Evaluation was accomplished through recorded semistructured interviews and quantitative surveys to capture demographic information and measure the system usability score. Interviews were transcribed and analyzed iteratively using NVivo (version 12; QSR International).

Results: This study included 8 caregivers (aged 33-50 years) of children with cancer, with most children receiving chemotherapy, and 6 nurse coordinators, with 3 (50%) of them having 11 to 20 years of nursing experience. The mean system usability score given by caregivers was 89.4 (95% CI 80-98.8). Results were grouped by app function assessed with focus on specific attributes that were well received and those that required refinement. The major issues requiring refinement included clarity in the medical information and terminology, improvement in design of tasks, tracking of symptoms including adjusting the look and feel of certain buttons, and changing the visual graph used to monitor symptoms to include date anchors.

Conclusions: The Cope 360 app was well received by caregivers of children with cancer but requires further refinement for clarity and visual representation. After refinement, testing among caregivers in a real-world environment is needed to finalize the Cope 360 app before its implementation in a randomized controlled trial.

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KEYWORDS

child; adolescent; oncology; supportive care; mHealth; mobile health; cancer; pediatrics; children; digital health; health applications; parent; caregiver

Introduction

Background

When a child is diagnosed with cancer, it is a life-altering event for both the patient and their caregivers [1]. After a new diagnosis, caregivers take on the immense burden of learning to navigate the health care system and provide at-home medical management. Although pediatric oncology providers play an important role in medical care, it is the caregivers who take on the burden of the hands-on, day-to-day care of the child with cancer. These roles of the caregiver can include the providing direct care, administering medication, assisting in activities of daily living, coordinating complex health care services, and providing emotional support [1-3]. Owing to the fact that many children with cancer had few serious medical needs before diagnosis, the weight of handling the new care demands can lead caregivers to experience distressful emotions, physical stress, and negative behavioral and physiological impacts [4-6]. A means by which we can improve caregiver outcomes could be to support their caregiving needs for their child with cancer.

Mobile health (mHealth), defined as the application of mobile or wireless communication technologies to health and health care [7], has tremendous potential to support caregivers in the medical management of their child with cancer. mHealth apps have been used successfully to support both patients and caregivers of adult patients with cancer [8-11]; however, none have been directly aimed at caregivers of children with cancer. In our recent investigation, we found that caregivers of children with cancer desired an mHealth app that would help them with the medical management of their child, specifically including medical knowledge, symptom tracking and management, and timely and convenient medication reminders [12]. These tracking and monitoring components of medical management could aid caregivers across the spectrum of their caregiving experience, including supporting them in the home setting, communicating with their oncology team about specific symptoms or concerns, and improving their preparedness when seeking urgent evaluation for a complication. In addition, preparing for potential medical emergencies is integral to caregiving for a child with cancer. Previous research has demonstrated that approximately half of the children with cancer will seek emergency department (ED) care within the first year after diagnosis [13]. Through our explorations of the experience of children with cancer and their caregivers when medical emergencies arose in the community setting, we found that the key components for emergency preparedness included the ability to easily connect with the oncology team, having a packing checklist, and an informational card to show the ED staff [14,15].

Objectives

The objective of this study is to collaborate directly with key stakeholders, including caregivers of children with cancer and oncology providers, to place them at the center of the design and development process of an mHealth app to support caregivers in the medical management of their child with cancer. The hypothesis is that input from end users will lead to further and necessary refinements before implementing this app in a real-world setting.

Methods

Study Design

This is a pilot, mixed methods research study to engage directly with end users (ie, caregivers of children with cancer) and proxies (ie, nurse coordinators who triage sick calls) to co-design and create an app to support caregivers in the medical management of their child with cancer. There were two phases in this project: walking through prototyping of the app (phase 1), followed by alpha testing directly with caregivers (phase 2). First, we describe the intended functions of the mHealth app and its features, and then explain phases 1 and 2 of our study.

Intended Functions of the mHealth App

Overview

Our team strived to create an app that combined the features previously documented as desirable and functional for caregivers of children with cancer [12]. These desired features included medical management features such as medical knowledge, symptom tracking, and medication reminders [12]. Medical knowledge could consist of specific details about the child's diagnosis, type of central line, and clinical recommendations for specific symptoms. The symptoms to track were based on literature related to the most common types of symptoms experienced by children with cancer, including pain, nausea and vomiting, diarrhea or constipation, fevers, and signs of breathing difficulties [16-20]. Medication reminders were created for both scheduled medications and supportive care medications if requested by the caregiver. It was also determined to be important to include a feature that aided caregivers in preparing to seek ED care for medical issues. The overall intent of the app was to assist caregivers in the medical management aspects of their child with cancer needs while they are in a home- or community-based setting. It was not intended to be used while patients were actively being evaluated by a medical professional or under the direct care of an oncologist (such as during hospital admissions for chemotherapy). Therefore, the app has three key functions: (1) patient information and caregiver team, (2) symptom tracking, and (3) emergency preparedness. Screenshots of the key screens are shown in Figure 1.

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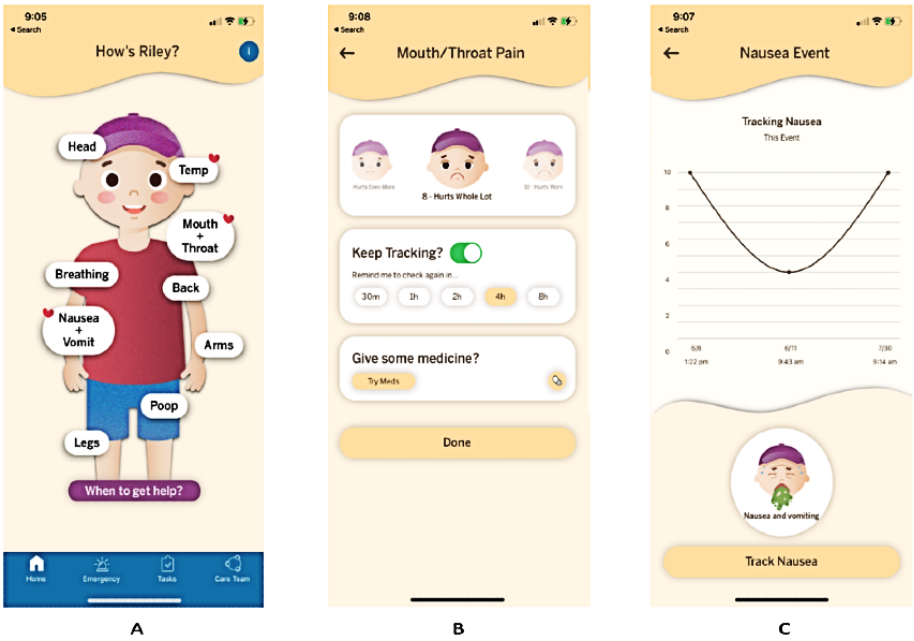
Figure 1. Images from the Cope 360 app including the (A) home screen view, (B) screen for documenting a symptom, and (C) screen for viewing the tracking of a symptom.



Patient Information and Caregiver Team

Patient information and caregiver team is where caregivers can add or view information on members of the caregiving team for their child. Under patient information and caregiver team, there is an open space to list the patient's nickname; drop-down menus for listing the patient's medical team based on our institution's practices; and a toggle for if the patient has a central line, which then leads to a drop-down for line type. On the caregiver team screen, the primary caregiver can type their name, use a drop-down to characterize their relationship to the patient, determine which types of notifications they would like to receive, and upload a photo for their profile. On this screen, the primary caregiver can also invite other caregiver team members through a link to their phone contacts list.

Symptom Tracking

The purpose of symptom tracking is to assist caregivers in tracking common symptoms experienced by children with cancer, identified based on previous literature [16-20]. The symptom tracking feature is located on the home screen, where there is a cartoon representation of the patient that can be personalized by gender and 3 skin colors. There are bubbles for nine areas of symptom tracking, including head, temperature, mouth and throat, breathing, back, arms, nausea and vomit, poop, and legs. There is also a link to *When to get help?* on the home screen. Each symptom has an individualized tracking scale based on previously published or validated scales. We used the Faces Pain scale for head, back, arms, and legs [21]. The Baxter Retching Faces scale was used for tracking nausea and vomit [22] and the Bristol scale was used for monitoring poop [23]. The temperature tracking provides direct feedback based on the temperature input from the caregiver. The *When to get help?* screen includes reasons to call 911 with a direct link or reasons to speak with someone from the pediatric oncology team with a direct link to the clinic or after-hours services based on the day and time.

Medication Reminders

Either the oncologist or the nurse coordinator enters the patient's current medications including scheduled medications and supportive care medications through the web-based application. Then, these are updated in the caregiver app, which will create reminders for scheduled medications. Once a symptom is tracked, the caregiver can also request reminders to administer supportive care medications until the symptom is no longer tracked.

Emergency Preparedness

The emergency preparedness plan screen allows the caregiver to create, practice, and enact a plan for seeking care for an urgent medical issue. The emergency preparedness plan screen will enable caregivers to pick their preferred ED and set up a contact plan with prescripted texts or a contact list to call and will provide a checklist of things to do, a packing list for items to bring, and finally, a *when you arrive* screen that can be shown to the ED staff.

Phase 1: Development and Rapid Refinement With Proxy Users

On the basis of previously published research on prototyping an mHealth app for children's oncology emergency planning, we learned that caregivers desire the ability to track symptoms and have medication reminders [12,15]. Therefore, we used these data to create the initial prototype, and then, we sought formative input from proxy users (ie, nurses) before initiating alpha testing with end users (ie, caregivers). We conducted rapid design interviews with nurse coordinators in our hospital system, who are health care professionals engaged in phone management and triaging of children with cancer who are experiencing medical emergencies in the home setting. At our institution, 7 nurse coordinators play this role. Demographic information, including age, gender, race, ethnicity, zip code, years of experience category, degree, and job role, was collected from

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the nurse participants. Nurse coordinator interviews were conducted using quick-and-dirty prototyping design methods by Buley [24], intended for proxy users of the final product. These interviews were conducted in person with the research team observing the nurse coordinator going through a series of tasks, including downloading the app, creating a profile, developing an emergency action plan, and opening and tracking each type of symptom (pain, nausea and vomiting, pooping, breathing, and fever). The nurse coordinators were observed for how often they encountered errors or if there was confusion with the intended function. They were encouraged to *think aloud* during the process, and the research team took notes [25,26]. The prototype was refined based on feasibility feedback provided by the nurse coordinators. These rapid prototyping sessions resulted in refinements to the app version in preparation for alpha testing with end users and proxies.

Phase 2: Alpha Testing With End Users

In phase 2 of the project, we used alpha testing to refine the app with caregivers of children with cancer. First, demographic information from the caregivers was collected using a web-based survey, including relationship to the child with cancer, age, gender, race, ethnicity, zip code, marital status, annual household income, and education. Then, alpha testing was accomplished through an audio-recorded semistructured qualitative interview and a quantitative web-based survey. For the interview, participants were asked to perform a series of tasks to test the usability of the prototype using the same series of tasks as the nurse coordinators: downloading the app, creating a profile, developing an emergency action plan, and opening and tracking each type of symptom (pain, nausea and vomiting, pooping, breathing, and fever). They were encouraged to *think aloud* [26] and comment or ask questions as they moved through the app. Then, the interviewer would follow up to probe deeper into the comment or to obtain clarification. The interviews were audio and video recorded so that during analysis, the reviewer could see which screen was being referenced. At the completion of the interview, caregivers completed a web-based survey using the System Usability Scale (SUS) for the app [27,28].

Collaboration and Ethics

Development and prototyping of the app were made possible through a partnership with Coactive Business Solutions of Indianapolis, Indiana. The Indiana University Institutional Review Board approved this study (number 1903250567).

Study Population and Identification of Cases

A convenience sample of pediatric oncology nurse coordinators employed at Riley Hospital for Children was used for testing among health care providers in phase 1. Nurse coordinators were contacted via email and scheduled for an in-person interview during their typical workday. In phase 2, the participants were caregivers of a child with cancer (the child had to be aged <21 years), had adequate English language proficiency with grossly normal cognitive function, and had a child who was currently receiving cancer therapy at Riley Hospital for Children and at least 1 month had passed after initial diagnosis. Nurse coordinator interviews were conducted both in person and via Zoom videoconferencing. Caregivers were contacted by phone to schedule the interviews, which were conducted and recorded over Zoom videoconferencing owing to COVID-19 restrictions.

Analyses

The research team created an initial codebook based on the series of tasks requested to be completed by each participant. For each task, codes were created for positive and negative comments. We conducted iterative thematic analysis on transcripts and notes from each interview. In each phase, interviews were conducted with participants (nurse coordinators in phase 1 and caregivers in phase 2) until no new information was gathered and thematic saturation was achieved [29,30]. Caregiver semistructured interviews were transcribed by a Health Insurance Portability and Accountability Act–compliant service and then analyzed using NVivo (version 12; QSR International) by three team members (MEC, ARC, and ELM). First, two team members (MEC and ARC) independently reviewed each transcript and assigned codes based on themes using an initial codebook based on the tasks that caregivers were asked to complete and comment on. Codes were revised based on new themes that emerged through data review [29,30]. A final review was performed with three team members (MEC, ARC, and ELM) until agreement on codes and themes was obtained. Findings from the transcripts were then grouped by similarity to create overarching themes. Data are presented as both features that worked well and those recommended for improvements in future versions of the app.

To evaluate usability, we chose to use the SUS [27,28], which has 10 questions on a 5-point Likert scale. The SUS has a calculated final score that is based on a well-established reference standard and is suitable for use even among small populations. A high SUS score indicates better product usability by the participants who evaluated it.

Results

Demographic Information of Phase 1 and Phase 2 Participants

A total of 6 nurse coordinators were interviewed in phase 1 of the prototype testing. Interviews lasted approximately 15 minutes on average. As presented in Table 1, all the nurse coordinators were women and White and non-Hispanic (6/6, 100%) and all of them had a Bachelor of Science in Nursing degree (6/6, 100%). Age ranged from 34-51 years, with a median age of 35 years. Job experience ranged from 3-5 years to ≥20 years and half of them (3/6, 50%) stated that they had 11-20 years of experience.

A total of 8 caregivers were interviewed for phase 2 of the prototype testing. Interviews lasted for approximately 25 minutes on average. As presented in Table 1, all caregivers were women, White, non-Hispanic, and parent-type caregivers (8/8, 100%). Half of the caregivers (4/8, 50%) had a child diagnosed with acute lymphoblastic leukemia, whereas the other half (4/8, 50%) had a child diagnosed with solid tumor type of cancer. Of the 8 children, 4 (50%) of them were undergoing chemotherapy only, whereas 3 (38%) were being treated with both chemotherapy and radiation and 1 (13%) was undergoing

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another type of treatment. Nearly all the caregivers were married (7/8, 88%), with 13% (1/8) of them being divorced. Most caregivers reported yearly household income of >US $75,000 and education level of college graduate or higher.

Table 1. Demographic characteristics of the participants alpha testing the Cope 360 app (N=14).

Characteristics	Caregivers (n=8)	Nurse coordinators (n=6)
Gender, n (%)		
Men	0 (0)	0 (0)
Women	8 (100)	6 (100)
Age (years; n=7), median (range)	40 (33-50)	35 (34-51)
Race and ethnicity, n (%)		
White, non-Hispanic	8 (100)	6 (100)
Type of cancer, n (%)		N/A[a]
Acute lymphoblastic leukemia	4 (50)	
Solid tumor	4 (50)	
Type of therapy, n (%)		N/A
Chemotherapy only	4 (50)	
Chemotherapy and radiation	3 (38)	
Other	1 (13)	
Type of caregiver, n (%)		N/A
Parent	8 (100)	
Marital status, n (%)		N/A
Married	7 (88)	
Divorced	1 (13)	
Yearly household income (US $), n (%)		N/A
<25,000	0 (0)	
25,000-49,999	1 (13)	
50,000-74,999	1 (13)	
75,000-99,999	2 (25)	
100,000-150,000	1 (13)	
>150,000	3 (38)	
Education, n (%)		
Less than high school	0 (0)	0 (0)
High school or GED[b]	1 (13)	0 (0)
Some college	2 (25)	0 (0)
College graduate	3 (38)	6 (100)
Graduate degree	2 (25)	0 (0)
Job experience (years), n (%)	N/A	
0-2		0 (0)
3-5		1 (17)
5-10		1 (17)
11-20		3 (50)
≥20		1 (17)

[a]N/A: not applicable.

[b]GED: General Educational Development.

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System Usability Score of Phase 2 Participants

When we evaluated the 8 caregivers' SUS responses, we found a mean score of 89.4 (95% CI 80-98.8). This falls above the generally recognized lower limit of acceptability for technology applications (≥70) [27,28].

Qualitative Exploration of Phase 2 (Caregivers) Interviews

For the qualitative evaluation of the caregivers' experience with the app during alpha testing in the *lab-based* setting, responses were grouped by app function with common themes of either positive attributes or future areas for refinement presented. Representative quotes of caregivers are shown in Table 2.

Table 2. Key quotes from caregiver interviews, grouped by theme.

Theme and function	Key quotes
App setup and planning	
App log-in and caregiver team creation	• "This is nice, select from contacts. Okay, search. That part's really nice." • "I know what a central line is because I'm a surgeon, but I'm not sure other people would know that."
Emergency action planning	• "Yeah. It just goes right to my contacts and pulled it through, so very easy. So, I added that."
Task list and planning list	• "Actually, I would probably add to this grabbing her medications only because the last time we went to the ER, we forgot them. Oh, my goodness sakes. We've been doing this for how long, and then we forgot it." • "I really like that because when you're in that moment, it's hard to remember everything...Yeah, that's cool." • "This is helpful because I feel like I always forget something..."
Seeking emergency care	
When you arrive	• "Yeah, and we have utilized two emergency room departments,...and both of them have been disastrous. So, just as I'm reading this, I'm like, oh my gosh. If I had something like this, I could be like, look, this is what has to happen. I think it would be huge." • "The first time we went to [a local hospital], the doctor was looking at me like, what do you want me to do?...So, I guess that would have been helpful to be able to show that to him like this is what their plan, what they recommend."
Medical management of care	
Logging symptoms	• "I think it's a very convenient, very easy, very helpful because we may not write down as much as what we should, and this would be very easy to just pull up and push the buttons and say okay, this is what's going on." • "Yeah. I mean, I think it would be better with words under it..."
Medications	• "Right, but it's cool though to have a listing of her medications. I don't know what type of information...I think [it would be helpful to have] because I know we get the papers, clinic or an inpatient, but I know those hardcopies just get sort of lost in the shuffle." • "I would say having one app where you manage everything, including the regularly scheduled meds, which are really honestly extremely important. That's the treatment."
Miscellaneous	• To-do list: "Yeah, and medicines would be good on there. Right now, I do it on a board, but it would be easier to do it in my phone, so I had it when I got over to the hospital."

App Setup and Planning

App Log-in and Caregiver Team Creation

First, caregivers were asked to log in to the app using their phone number and were provided an access code through an SMS text message. All participants were able to successfully enter the app; however, 13% (1/8) of them had difficulty in receiving the access code but eventually received it. Next, they were asked to create an account. Almost all (7/8, 88%) of them commented that they were able to add other caregivers to the app with no or few difficulties. Of the 8 caregivers, 1 (13%) caregiver did not comment on whether they had any difficulties regarding this. A few caregivers (3/8, 38%) were initially confused if a port-a-cath (port) was considered a central line when adding patient information to create an account. However, once the central line was selected, they were able to select the port from a drop-down list of line types and understood the setup. Overall, caregivers were able to easily set up an account in the app with little to no assistance from the interviewer; however, not all of them commented on it.

Emergency Action Planning

Then, the caregivers were asked to set up an emergency action plan, which had several components. They were asked for their preferred ED, their contact plan (a place where they can set up SMS text messages or phone calls to other people if they are going to the ED); a *before you leave* section, where they can be

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reminded of tasks that need to be done before leaving for the ED; a packing list; a *when you arrive* section, which contained general instructions for the treatment of a child with cancer that can be shared with the ED; and finally, a *when to get help* informational section. Overall, the caregivers found it easy to set up an emergency action plan. Caregivers who added other people from their contact list to their emergency action plan said that it was easy to do. They also found it easy to edit SMS text messages when asked how they would do it by the interviewer.

Task List and Packing List

All caregivers appreciated the task list that included several prepopulated tasks including to call the oncology team and pack. There were additional tasks that caregivers could add to their task list including bringing home medications, packing the wheelchair, and seeking childcare for other children at home. When asked to add items to the packing list, many caregivers thought they had to press the plus button and then start typing instead of typing and then pressing the plus button. After they understood the correct method, they stated that it was easy to do. The caregivers had positive thoughts on the packing list with examples of items they could add, including laptops, medications, food, extra clothing, toiletries, and so on.

Seeking Emergency Care

When to Get Help

Caregivers reviewed the information contained in the *when to get help* section of the emergency action plan. Almost all caregivers (7/8, 88%) commented that the information presented in this section is clear. Specifically, they appreciated the capacity to call directly from the app if their child was experiencing a serious symptom. The addition that 13% (1/8) of the participants requested was for *uncontrolled pain* to be added as a reason to seek care.

When You Arrive

When asked to examine the information in the *when you arrive* section, all caregivers stated that the information would be helpful if they ever had to go to an ED outside of their treating institution. Caregivers stated that the information was very useful to explain general details about what the child is going through. A caregiver stated that defining what a fever is for a child with cancer would be helpful to add to the card, and another caregiver thought that port needle size was important to be included.

Medical Management of Care

Logging Symptoms

Caregivers were asked to log a series of commonly monitored symptoms, which included fever, pain, poop, nausea and vomiting, and breathing. All caregivers were able to log symptoms successfully. Once a symptom was logged, they were asked if they wanted to continue tracking and at what time intervals they wanted to be reminded to check again. They also had the option to set medication reminders for certain medicines as needed. Caregivers were told by the interviewer that medications and dosages available for their symptoms would be entered by the medical team; however, this aspect was not available for this phase of testing. Then, the caregivers were asked to go through and track each symptom and were asked if tracking of each symptom was clear and easy to do. For example, caregivers said that it was easy to track headache, and they appreciated the different faces showing levels of pain or discomfort.

Some caregivers stated that a description under different nausea and vomiting faces would be helpful, as some symptoms such as nausea and vomiting and pooping had only a number identifier, unlike pain, which included a number scale and a descriptor of the level of pain. Of the 8 caregivers, 5 (63%) caregivers thought that the poop scale was not self-explanatory. A description of each type or definition of *normal* would make it easier to gauge.

All caregivers (8/8, 100%) said that tracking the temperature was clear and easy to do; however, 13% (1/8) of the caregivers was initially confused at the initiation of the emergency action plan when tracking a dangerously high temperature. Some comments were provided on the graphing ability of temperature symptom tracking to be able to view the tracking of multiple temperature readings over time.

Medications

Although medications were not available for review by the caregivers during alpha testing, the *give medications* option was shown during symptom tracking, and caregivers were asked about their thoughts on using the app for medication tracking and reminders. Overall, the caregivers were supportive of using the app for this purpose. However, 13% (1/8) of the caregivers expressed concern over whether they would trust the medication information contained within the app.

Completing a Symptom Tracking Event

Caregivers were asked how they would end a tracking event if they no longer wanted to track a symptom. Of the 8 caregivers, 5 (63%) caregivers did not have any problems in understanding how to end the tracking. The remaining 38% (3/8) of the caregivers needed to be guided through the process, and 33% (1/3) of them said that it was clear after they were shown what to do. Improved ease and clarity are needed in how to complete a tracking event.

Miscellaneous App Functions

Pulsing Heart Perceptions

Caregivers were asked what they thought the *pulsing hearts* on the home screen meant after they tracked a symptom—more than one symptom at a time can have a *pulsing heart*. Most caregivers (7/8, 88%) knew that the heart meant that they were tracking that particular symptom. Only 13% (1/8) of the caregivers thought that the pulsing heart meant that the symptom being tracked was *good*.

To-do List Section

All reminders that caregivers have set show up in the *care tasks* section. Caregivers were asked if there was anything else that they would like to see in this section. Multiple caregivers

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mentioned that they would like to see medication (7/8, 88%) and appointment reminders (4/8, 50%) in this section.

Overall Impression

When asked if there were any final comments or concerns, several caregivers expressed their overall satisfaction with the app and could envision that it would serve a meaningful purpose to caregivers of children with cancer. Several key quotes from caregivers included the following:

> *I mean, it seems like you guys have everything on here right now that we've ran into.*
>
> *I just want to tell you guys kudos because this is very self-explanatory. I feel like it's very easy to follow and to understand...It's user friendly, and I don't feel intimidated by this program. I'm like oh, this is really awesome, and this makes good sense, and it walks me through if ever there's a time where I'm questioning it pretty well, once I put it in there, it's telling me yeah, you need to be calling the doctor.*
>
> *It looks good. We're kind of 30 weeks in, but at the beginning to have all that information available would be very helpful, for sure.*

Discussion

Principal Findings

In this mixed methods study, we document the process and importance of involving key stakeholders in the prototyping and alpha testing of an mHealth app to support caregivers in the medical management of their child with cancer. The use of an app, such as Cope 360, that has been co-designed and created with input from the intended users has the potential to positively impact caregiver outcomes. Overall, the app was well accepted by caregivers of children with cancer, but several key issues arose that require refinement before further studies. Before we can explore the impact of this app, future work will need to be conducted to explore user experience and preferences in the real-world setting.

On the basis of the results of the qualitative exploration of the interviews with caregivers of children with cancer, several key refinements will be made to the Cope 360 app. We describe these based on two categories: medical information and terminology and more clarity in design and features to be included in the beta phase of testing. Then, we explain some next steps and the importance of human-centered design when designing mHealth apps to assist caregivers.

Medical Information and Terminology

We found the importance of using proper, clear, and consistent terminology for medical terms. For example, the terminology regarding the type of line given to a particular patient was not universally well understood by caregivers, and the wording of the line type needed to change to *tunneled central line* instead of *central venous catheter*. We also learned about the importance of including more details concerning the patient's medical information. For example, it was important to allow the addition of the size of the port needle to be part of the patient information in the app. On the *when you arrive* section to show to the emergency providers, caregivers also desired that the definition of fever be added, as this is a more complicated and specific definition than that used in general pediatrics [31].

Improved Clarity in Design and Features to Be Included

Caregivers needed more clarity in the design of specific features to facilitate use in general and increase understanding for the first-time user. In the areas where the caregiver could add tasks or items to a list, there was confusion about the location and intent of the plus button; therefore, this was recommended to change to only show the plus button on the left-hand side of the screen with the option for typing content after the plus button was pressed. For symptom tracking, caregivers requested more consistency between the types of symptom tracking with the addition of descriptors along with numerical representation for nausea and vomiting. The Bristol poop scale was not easily understood, and caregivers desired an option for *no poop* when their child attempted but was unable to poop. Therefore, refinement to the poop scale was made with descriptors and an option for *no poop*. All symptom tracking charts were updated to include the date the symptom was tracked along with the time. These recommendations were relayed to the app development company.

Future Steps and Directions

Very often, the intended end users are not included in the up-front design and creation of the mobile technology app [32-34]. This often results in the end product not aligning with the experiences and needs of the end user, which ultimately leads to poor uptake of the app or its incorporation into daily life. For this app, we leaned on the lived experience of caregivers of children with cancer to understand the intricacies of their experience with medical management of their child. This helped us identify important gaps in the mHealth app and quickly identify ways in which we could adjust small features to better accommodate the caregivers' desires and needs. Our goal is that by creating an mHealth app with caregivers and for caregivers, we will be able to positively impact their experience with providing medical care to their child with cancer. The Van Houtven Caregiver Intervention Organizing Framework suggests that interventions to improve a caregiver's clinical skills and knowledge, psychosocial (self-efficacy and coping) competency, support seeking (organizational and coordination), and quantity of caregiving will lead to benefits for both them and the patient with cancer [35]. For this project, we co-designed and created an app that is intended to improve caregivers' clinical skills and knowledge, self-efficacy, and support seeking skills.

Although we were successful in collaborating directly with caregivers of children with cancer, our success was also dependent on input from our nurses who acted as proxy users. The nurses included in this study provide phone triage to caregivers of children with cancer regularly as part of their roles as nurse coordinator. They have the advantage of understanding the clinical context of the symptoms that we are tracking and the communication needs of the medical team when helping the caregiver relay questions or concerns. The disadvantage may have been that the nurses approach symptom tracking and emergency preparedness as a daily activity, whereas this can

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be a new and daunting task for caregivers. By combining the nurse coordinators' inputs with testing by caregivers, we are hopeful that we can create a both practical and supportive app for the medical management of children with cancer.

The results from the alpha testing reported in this paper provide both encouraging signs that the app is likely to be useful and clear next steps for certain features and functionality. On the basis of these findings, we are planning a beta test with increased functionality and revised features and interface. Specific features that are anticipated to be explored further during beta testing include perceptions or alterations to the pulsing heart that signals an active symptom tracking, preferences for how to stop tracking an event, allotted time intervals for rechecking a symptom, and any additional recommendations for medical knowledge related to when to seek emergency care. Our intent is to use the mHealth app as part of a randomized controlled trial that specifically measures caregiver outcomes including mastery of caregiving, caregiver self-efficacy, and stress [36-39].

Limitations

All the participants in this study, including proxy users (ie, nurse coordinators) and caregivers, were recruited from a single institution. There was a lack of male participants and those with race and ethnic backgrounds other than White, non-Hispanic. However, we received varying opinions on certain features and were able to collect data until no further themes emerged. Of note, alpha testing began when the COVID-19 pandemic changed the way in which we interacted with patients and research participants. The research team transitioned all recruitment, interview, and evaluation processes to a web-based platform. Although the interviews were audio recorded and transcribed, the opportunity to observe in more detail how the caregivers interacted with the app was hindered.

Conclusions

By using a mixed methods approach for prototyping and alpha testing, we were able to create and refine an app to support caregivers in the medical management of their child with cancer. By placing the intended users at the forefront of the app design process, we created an app that was well received by caregivers of children with cancer. However, some key features require refinements based on collective feedback. Future research should focus on how caregivers can use this app in real life to manage the medical needs of their child with cancer. Refinements after real-life testing would allow large trials to evaluate the impact of this app on caregiver outcomes, such as caregiver's feeling of self-efficacy and mastery of caregiving.

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Conflicts of Interest

None declared.

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Abbreviations

ED: emergency department
mHealth: mobile health
SUS: System Usability Scale

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Original Paper

Development and Evaluation of the Usefulness, Usability, and Feasibility of iNNOV Breast Cancer: Mixed Methods Study

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Abstract

Background: Despite the efficacy of psychosocial interventions in minimizing psychosocial morbidity in breast cancer survivors (BCSs), intervention delivery across survivorship is limited by physical, organizational, and attitudinal barriers, which contribute to a mental health care treatment gap in cancer settings.

Objective: The aim of this study is to develop iNNOV Breast Cancer (iNNOVBC), a guided, internet-delivered, individually tailored, acceptance and commitment therapy–influenced cognitive behavioral intervention program aiming to treat mild to moderate anxiety and depression in BCSs as well as to improve fatigue, insomnia, sexual dysfunction, and health-related quality of life in this group. This study also aims to evaluate the usefulness, usability, and preliminary feasibility of iNNOVBC.

Methods: iNNOVBC was developed using a user-centered design approach involving its primary and secondary end users, that is, BCSs (11/24, 46%) and mental health professionals (13/24, 54%). We used mixed methods, namely in-depth semistructured interviews, laboratory-based usability tests, short-term field trials, and surveys, to assess iNNOVBC's usefulness, usability, and preliminary feasibility among these target users. Descriptive statistics were used to characterize the study sample, evaluate performance data, and assess survey responses. Qualitative data were recorded, transcribed verbatim, and thematically analyzed.

Results: Overall, participants considered iNNOVBC highly useful, with most participants reporting on the pertinence of its scope, the digital format, the relevant content, and the appropriate features. However, various usability issues were identified, and participants suggested that the program should be refined by simplifying navigation paths, using a more dynamic color scheme, including more icons and images, displaying information in different formats and versions, and developing smartphone and tablet versions. In addition, participants suggested that tables should be converted into plain textboxes and data visualization dashboards should be included to facilitate the tracking of progress. The possibility of using iNNOVBC in a flexible manner, tailoring it according to BCSs' changing needs and along the cancer care continuum, was another suggestion that was identified.

Conclusions: The study results suggest that iNNOVBC is considered useful by both BCSs and mental health professionals, configuring a promising point-of-need solution to bridge the psychological supportive care gap experienced by BCSs across the survivorship trajectory. We believe that our results may be applicable to other similar programs. However, to fulfill their full supportive role, such programs should be comprehensive, highly usable, and tailorable and must adopt a flexible yet integrated structure capable of evolving in accordance with survivors' changing needs and the cancer continuum.

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KEYWORDS

acceptance and commitment therapy; anxiety; breast cancer survivors; cognitive behavioral therapy; depression; digital mental health; e-mental health; user-centered design; internet interventions; usability; mobile phone

Introduction

Background

Since 2020, breast cancer has been the most diagnosed cancer worldwide and the leading cause of cancer mortality in women. In Portugal, as many as 7041 women are diagnosed per year with breast cancer, and in 2020, a total of 1864 women died owing to the condition [1,2]. Nevertheless, owing to improvements in early diagnosis, tumor molecular characterization, and innovative systemic treatments, breast cancer prognosis has significantly improved across the globe, with 5-year survival rates reaching approximately 90% in high-income countries [3]. In Portugal, the 5-year prevalence of breast cancer was estimated at 27,051 in 2020, making breast cancer survivors (BCSs) the largest group of cancer survivors in the country [1,2].

In spite of a positive prognosis, the survivorship trajectory is frequently characterized by difficulties associated with sequelae of cancer and its treatment and late physical and psychosocial effects that hinder BCSs' health-related quality of life (HRQoL) [4]. Anxiety, depression [5,6], fear of recurrence [7], fatigue [8], sleeping problems [9], and sexual dysfunction [10,11] are among the most common problems BCSs experience across survivorship and can manifest up to several years after primary treatment completion [12].

In the past decades, several interventions have been developed to minimize psychosocial morbidity in BCSs. Recent meta-analyses demonstrated the efficacy of such interventions in improving a range of psychosocial outcomes [13,14]. Cognitive behavioral therapy (CBT) has been identified as the most effective intervention to treat anxiety and depression in BCSs, often showing significant small-to-moderate treatment effects in patients with these conditions [13,14]. Other psychosocial interventions such as psychoeducational treatments [14], mindfulness-based interventions [15], and acceptance and commitment therapy (ACT) have been tested among BCSs with success as well [16]. ACT, owing to its model of healthy adaptation to difficult circumstances and transdiagnostic approach, has been appointed as particularly useful in addressing the high levels of psychological and medical comorbidities that manifest in cancer populations [17,18]. Regrettably, the delivery of such interventions across the survivorship trajectory is limited owing to distance from health care services, health care system limitations, mental health illiteracy, and attitudinal barriers, all of which contribute to a mental health care treatment gap in cancer settings [19].

Internet interventions—self-help technology-enabled interventions that provide synchronous or asynchronous health-related and mental health–related assistance based on established psychotherapy models [20]—provide an opportunity to fulfill the mental health care gap within oncology and offer BCSs with patient-centered support at a distance. Nevertheless, despite internet interventions' attested efficacy [21] in treating various mental health conditions and its potential cost-effectiveness [22,23], internet interventions targeting cancer survivors are scant [24]. Although promising effects of such interventions have been documented concerning anxiety, depression [25,26], distress [27], fatigue [28], physical activity [29], symptom management [30], insomnia [31,32], sexual dysfunction [33], and quality of life [34,35], the overall benefit of such interventions for BCSs is still unclear. Most studies in this domain report on dissimilar interventions or present high methodological heterogeneity, which makes their comparison difficult and inconsistent [30,36]. Moreover, interventions' design processes are rarely reported, and the absence of evidence-based reasoning behind its development [36] contributes to a research-practice gap in the internet interventions domain, wherein evidenced-based treatments struggle to be adopted in routine care [37]. Another cause for the low uptake of internet interventions in clinical settings is the peripheral position end users are often referred to during development [38]. Intervention programs are frequently planned by neglecting end users' perspective (eg, individual's goals, needs, skills, and contexts) and researchers often fail to involve end users in the development process [37,38]. This lack of human-centeredness in the development partly explains the high attrition rates and poor engagement often reported in clinical trials and configures a limitation that needs to be addressed to effectively impact survivorship supportive care provision [39,40].

The aim of this study is to report on the development, usefulness, usability, and preliminary feasibility of iNNOV Breast Cancer (iNNOVBC), a guided, internet-delivered, individually tailored, ACT-influenced CBT program developed to treat mild to moderate anxiety and depression in BCSs, as well as to improve fatigue, insomnia, sexual dysfunction, and HRQoL in this group. Besides informing iNNOVBC's further development and refinement, the contribution of this paper resides in the description of mental health professionals' (MHPs') and BCSs' perspectives on the use of digital technology to support cancer survivors and the design implications that arise from considering these.

iNNOVBC Overview

iNNOVBC (Figures 1 and 2) is a guided, internet-delivered, individually tailored, ACT-influenced CBT program and was developed with a user-centered design approach [37]. The program was created to address the psychosocial needs of BCSs that were previously identified via literature review, namely, anxiety, depression, fatigue, insomnia, sexual dysfunction, and HRQoL. The intervention structure and content build up on prior CBT- and ACT-inspired interventions developed by the Department of Behavioural Sciences and Learning at Linköping University, targeting other populations [41-45]. The applicable

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previously available content was translated from Swedish and English to Portuguese and reviewed by external experts (ie, an oncologist, a nurse, a psychologist, and a BCS). Additional content was developed based on peer-reviewed sources [46-50], as well as mixed methods research conducted by the research team and involving iNNOVBC's primary and secondary end users [51,52].

iNNOVBC is composed of 10 treatment modules (Multimedia Appendix 1), namely, five mandatory modules (living with breast cancer and beyond, depression, anxiety, relaxation, and key points summary, and planning for the future) and five optional modules (behavioral activation parts I and II, sleep fatigue, and interpersonal relationships, sex, and intimacy), to be completed in 10 weeks. An introductory module provides general information about the program. Each module is designed to be completed in approximately 60 minutes and includes written text, images, videos, audio files, quizzes, ACT- and CBT-based exercises, homework assignments, and respective worksheets. The program adopts a transdiagnostic structure, featuring psychoeducation, acceptance, cognitive defusion, connecting with values, committed action, exposure, behavioral activation, and relaxation as central components. Sleep management, energy conservation, problem solving, and sensate focusing techniques are complementary components of the program. To guarantee optimal use of the program, the study intervention was developed according to the following persuasive system principles categorized by Oinas-Kukkonen and Harjumaa [53]: responsiveness, tunneling, tailoring, personalization, reminders, and professional support.

At the onset of the intervention, BCSs using the program should tailor their treatment with the support of their assigned therapist and according to their baseline assessment and preferences. Once they reach an agreement, the selected modules should be prescribed and made available weekly to the BCSs. Then, BCSs are prompted to complete the modules in approximately 1 week. Within 24 hours of module completion, the therapists assess the BCSs' progress based on the reported outcomes and determine whether they should proceed to the next module. When a new module is made available, BCSs receive an email notifying them. If not, therapists should instruct them on what needs to be completed to be able to advance to the next module. Integrated 2-way communication features such as email, chat, SMS text messaging, and videoconference support the intervention. The program is delivered via iTerapi, a web-based treatment platform developed at Linköping University [54].

Figure 1. The landing page of the iNNOV Breast Cancer program.

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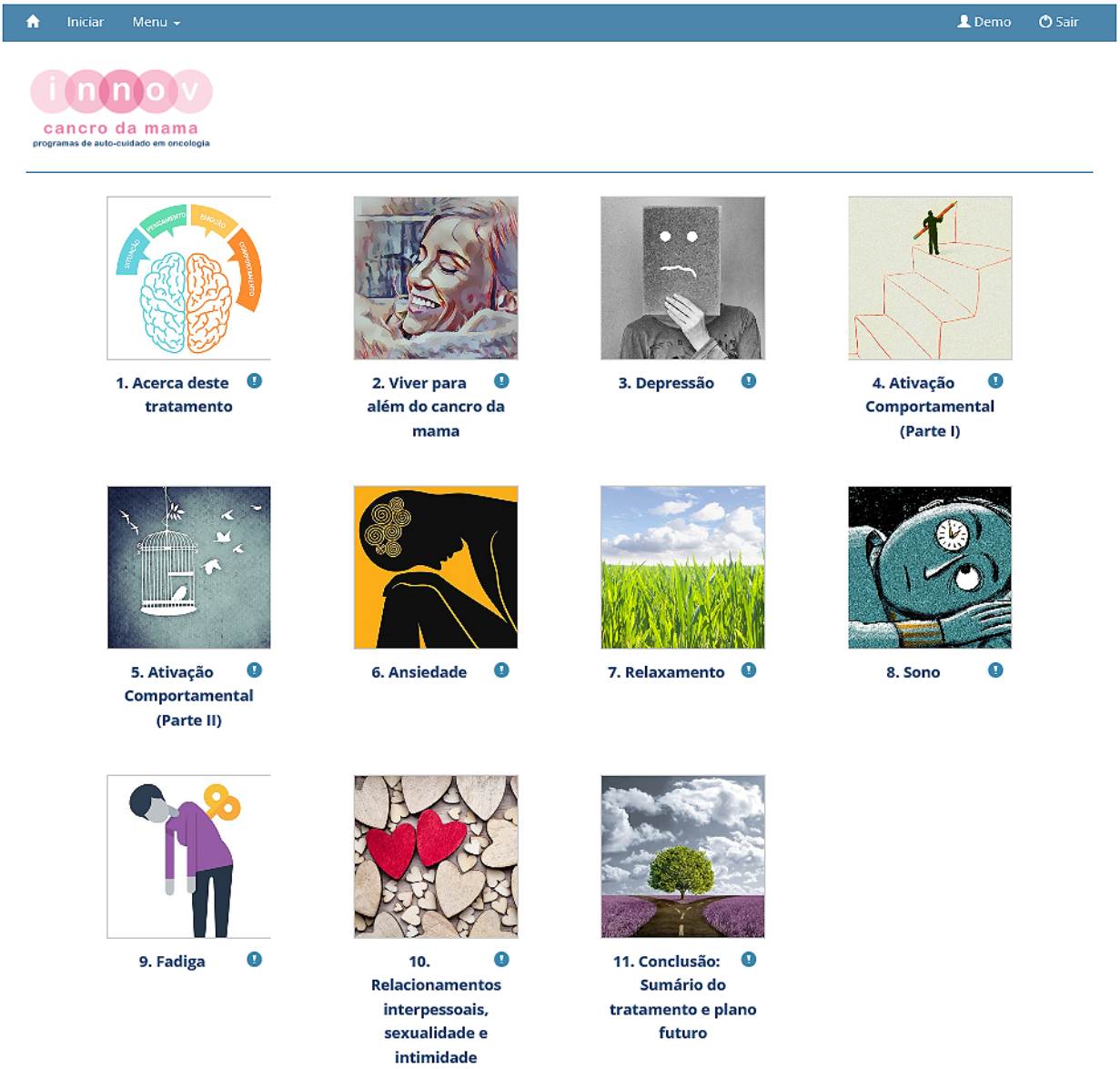
Figure 2. The introductory and 10 treatment modules of the iNNOV Breast Cancer program.



Methods

Study Design

Mixed methods (Figure 3), namely in-depth semistructured interviews, usability tests, short-term field trials, and surveys, were combined to fulfill the following specific goals: (1) evaluate iNNOVBC's usefulness, (2) assess iNNOVBC's usability, and (3) explore iNNOVBC's perceived feasibility and acceptability among its target users. The study was approved by the ethical committees of Instituto Português de Oncologia do Porto, Francisco Gentil, EPE; Centro Hospitalar Universitário do Porto; Centro Hospitalar São João; Unidade Local de saúde–Matosinhos; Hospital CUF Porto; Ordem dos Psicólogos Portugueses; and the Portuguese Data Protection Committee (approval number: 10727/2017). Written informed consent was obtained from all participants.

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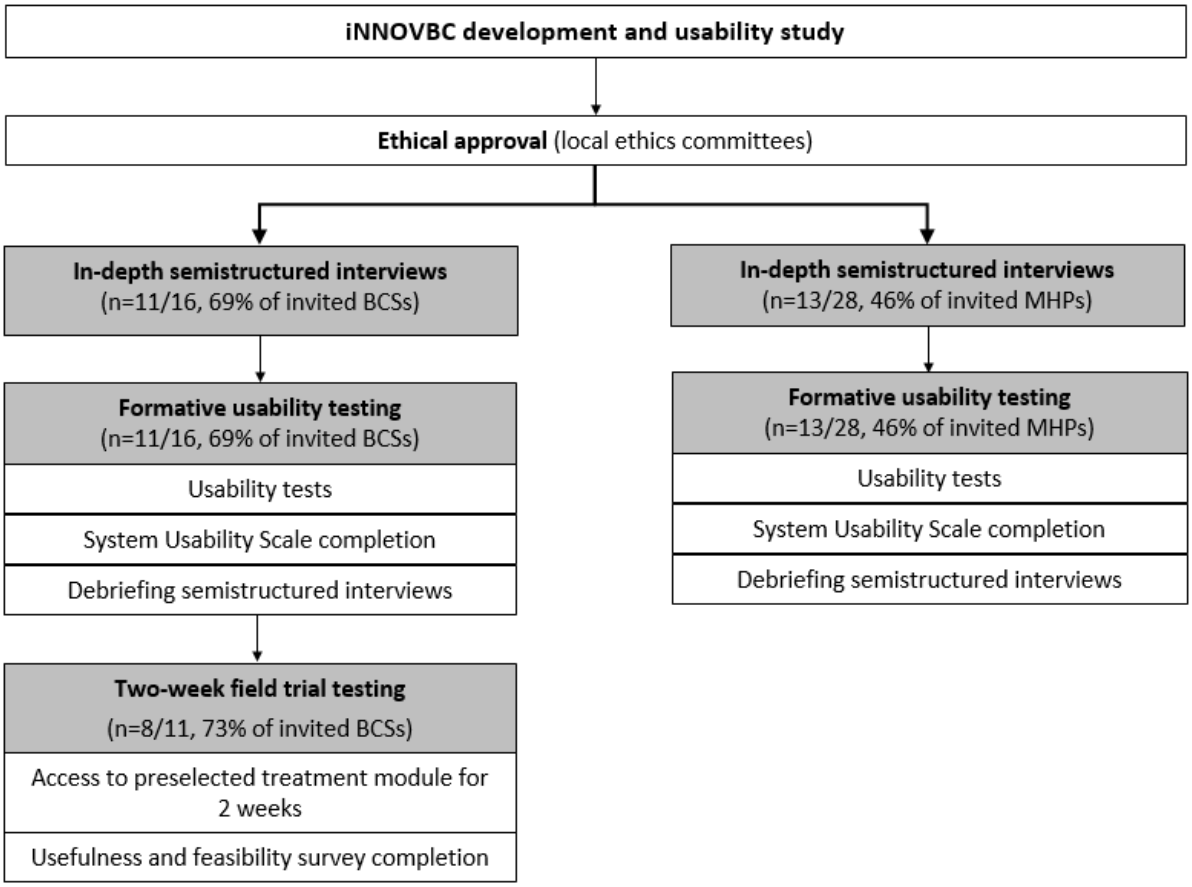
Figure 3. Study design. BCS: breast cancer survivor; iNNOVBC: iNNOV Breast Cancer; MHP: mental health professional.



Sampling and Recruitment

The study targeted BCSs and MHPs, who are iNNOVBC's primary and secondary end users. Eligibility criteria for BCSs consisted of women, aged >18 years, with a history of histologically confirmed breast cancer, who had completed primary adjuvant treatment (except hormonal therapy), and were capable of reading and writing in Portuguese. MHPs were registered psychologists or psychiatrists, capable of reading and writing in Portuguese. A nonprobabilistic sample of BCSs and MHPs was recruited following referrals from the researchers and professionals working at treatment centers in Porto (Portugal) and via snowball sampling. Participants were purposively sampled for diversity in age, academic degree, and digital technology proficiency. A total of 28 MHPs and 16 BCSs were invited to participate in the study in person, via email, or by telephone. Of the invited individuals, 54% (15/28) of the MHPs and 88% (14/16) of the BCSs agreed to participate. Meaning saturation was used as a stopping criterion, which meant that new participants would not be enrolled once novel fieldwork insights stopped changing analysis significantly [55]. Data collection ended after 87% (13/15) of the MHPs and 69% (11/16) of the BCSs completed the research protocol, as meaning saturation was reached during the last interviews and usability tests.

Data Collection Procedures

Data were collected between November 2019 and February 2020 (ie, before the COVID-19 pandemic). Up to 2 to 3 interviewers or moderators (CMS, Ana Alves, and Elsa Oliveira) participated in data collection. The research protocol included an exploratory semistructured interview (N=24; ie, 11/24, 46% BCSs and 13/24, 54% MHPs; Multimedia Appendix 2), a laboratory-based usability test (Multimedia Appendix 3), followed by the completion of a self-reported usability survey [56] (N=24; ie, 11/24, 46% BCSs, and 13/24, 54% MHPs), and a debriefing post usability test interview (N=24; ie, 11/24, 46% BCSs, and 13/24, 54% MHPs; Multimedia Appendix 4). The exploratory interview aimed to investigate the usefulness of iNNOVBC and gather requirements for further refinement of the program. The usability test assessed the participants' performance while executing a series of representative predefined tasks on distinct parts of the platform. The think-aloud protocol was implemented to enable participants to voice their thoughts and issues [57]. The debriefing interview focused on participants' experience with using the platform, issues hindering their experience, and changes to be performed to achieve better effectiveness, efficiency, and satisfaction with the program. These activities occurred in person at Fraunhofer Portugal–Centre for Assistive Information and Communication Solutions meeting rooms. Participant BCSs willing to further assess iNNOVBC were invited to participate in an additional 2-week field trial (8/11, 73% BCSs; Multimedia Appendix 5)

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mimicking the experience of using the program, and to fill out a survey through the platform on the perceived usefulness and feasibility of the program (Multimedia Appendix 6).

Interviews and usability tests were audio and video recorded, respectively. Approximately 36 hours of audio recordings and 9 hours of video recordings were created. The average duration of pretest interviews was 49 minutes (range 26-81 minutes) for MHPs and 74 minutes for BCSs (range 39-106 minutes). Usability tests were completed on average in 20 minutes and followed by 28 minutes (range 10-46 minutes) of debriefing interviews. Interview recordings were transcribed verbatim (CMS and Ana Alves) in parallel to data collection, using oTranscribe (created by Elliot Bentley; a project of the MuckRock Foundation) [58]. Usability tests were assessed using predeveloped observation grids and by registering participants' voiced remarks. Data were stored in a pseudoanonymized format in a secure password-protected location.

Measures

Clinical, Sociodemographic, and Internet-Related Characteristics

Background questionnaires tailored to each target group collected clinical, sociodemographic, and internet-related characteristics. BCSs were inquired about age, gender, education level, marital status, occupation, professional status, distance between residence and treatment center, time since diagnosis, type of treatment performed, survivorship status, proficiency using digital technology, and experience in using digital mental health (DMH) programs. MHPs were inquired about age, gender, education level, marital status, occupation, professional status, work context, professional experience (in years), theoretical orientation, proficiency in using digital technology, and experience in using DMH programs.

Usefulness

Semistructured interview guides (Multimedia Appendix 2) were developed based on a literature review to assess participants' perceived usefulness of the program. The interview guides covered the following domains: (1) survivorship main challenges, unmet care needs, and self-care strategies developed to address those challenges and needs; (2) the provision of psychosocial survivorship care to BCSs and the main barriers impacting it; (3) knowledge and use of DMH; and (4) attitudes toward DMH programs aimed at providing survivorship support. In addition, a questionnaire developed to assess the quality of iNNOVBC's treatment modules was used for this purpose (Multimedia Appendix 6).

Usability

We conducted usability tests and analyzed the performance and acceptance of the system. To assess performance, task analysis was conducted and the number of completed tasks, errors, and assistances were recorded in observation grids (Multimedia Appendix 3). Acceptance was measured using the Portuguese version of the System Usability Scale [56], where a score <68 is considered below average. The debriefing interviews also informed about acceptance and contributed to identifying content and design changes to be performed to improve user experience and satisfaction (Multimedia Appendix 4).

Feasibility

The preliminary feasibility of the program was assessed via debriefing semistructured interviews and assessment questionnaire in iNNOVBC's treatment modules (Multimedia Appendix 6). The participant BCSs who used the system at home also provided written comments.

Analysis

A combination of quantitative and qualitative methods was applied to assess iNNOVBC's usefulness, usability, and perceived feasibility. Descriptive statistics, namely, counts, percentages, medians, and IQRs, were used to characterize the study sample, evaluate performance data, and assess preference data collected via the System Usability Scale and the treatment modules' assessment questionnaire. No efficiency metrics were computed because the think-aloud method was applied. Microsoft Excel was used to compute quantitative variables. Qualitative data resulting from the interviews and BCSs' written comments were transcribed verbatim and analyzed using the thematic analysis method of Braun and Clarke [59]. First, a deductive approach was adopted based on three predetermined high-level themes: usefulness, usability, and feasibility. Subsequently, inductive analysis was performed on the data collected within those themes, and salient subthemes were coded. Initial coding was performed by the first author (CM). Regular discussions were promoted between researchers (CMS and FN) to discuss results and coding trees. Data patterns were then identified and iteratively organized (CMS and FN) until consensus between researchers was achieved and no additional insights were resulting from the analysis of the data. Scrivener software [60] was used to support the coding process.

Results

Participants' Characteristics

The sample comprised 24 participants (Table 1), including 11 (46%) BCSs and 13 (54%) MHPs. The median age of BCSs was 48 years (IQR 14; minimum 32, maximum 68). Most survivors were married (7/11, 64%) and college-educated (7/11, 64%). Approximately 55% (6/11) of the survivors were professionally active. Most BCSs had been treated with surgery (11/11, 100%), chemotherapy (9/11, 82%), radiotherapy (8/11, 73%), and hormonal therapy (9/11, 82%). Considering the survivorship status [61], the sample was heterogeneous, including participants at acute (2/11, 18%), extended (3/11, 27%), and permanent (6/11, 55%) survivorship stages. No participant had previous experience in using DMH programs, but the majority reported medium (4/11, 36%) to strong (5/11, 45%) skills in using digital technology.

The subsample of MHPs was composed of 15% (2/13) psychiatrists and 85% (11/13) clinical psychologists. Most professionals were female (11/13, 85%), and their median age was 35 years (IQR 11; minimum 25, maximum 56). The median of MHPs' professional experience was 11 years (IQR 11). Moreover, 92% (12/13) of professionals were active, and 30% (4/13) worked in psycho-oncology services. Approximately

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half of our sample (7/13, 54%) held a CBT orientation. Considering participants' proficiency in using digital technology, most professionals (11/13, 85%) classified their skills as medium. Approximately half of professionals (7/13, 54%) reported previous experience in using DMH programs.

Table 1. Participants' characteristics (N=24).

Variables	MHP[a] (n=13)	BCS[b] (n=11)
Sex, n (%)		
Female	11 (85)	11 (100)
Male	2 (15)	0 (0)
Age (years), n (%)		
23-30	3 (23)	0 (0)
31-40	7 (54)	4 (36)
41-50	2 (15)	4 (36)
51-60	1 (8)	2 (18)
61-70	0 (0)	1 (9)
Highest academic degree, n (%)		
Basic degree (≤9 school years)	0 (0)	2 (18)
Secondary degree (12 school years)	0 (0)	2 (18)
University degree	13 (100)	7 (64)
Self-reported proficiency in using digital technology, n (%)		
Poor	1 (8)	2 (18)
Medium	11 (85)	4 (36)
Strong	1 (8)	5 (45)
Self-reported use of digital mental health, n (%)		
None	6 (46)	11 (100)
Occasional	4 (31)	0 (0)
Regular	3 (23)	0 (0)

[a]MHP: mental health professional.

[b]BCS: breast cancer survivor.

Usefulness

iNNOVBC's perceived usefulness was assessed during the interviews conducted before and after the usability tests and by surveying field trial participants on the usefulness and adequacy of the program's modules. During the field trials, the content of the modules living with breast cancer and beyond (4/11, 36%), anxiety (1/11, 9%), relaxation (1/11, 9%), sleep (1/11, 9%), and fatigue (1/11, 9%) were appraised, with all being rated with 5 (out of 5) stars by BCSs evaluating it. Overall, participants found the program's approach, content, and features as highly relevant and useful. BCSs classified the content of the modules as very useful (3/8, 36%) or extremely useful (5/8, 63%) and very adequate (3/8, 36%) or extremely adequate (4/8, 50%) in addressing their difficulties and needs. Only 13% (1/8) of the participants reported that the relaxation module was neither adequate nor inadequate to her needs. All survivors (8/8, 100%) reported that they would recommend the modules to a friend or family member going through the same condition.

Considering the role iNNOVBC could play in BCSs supportive care, both survivors and professionals considered iNNOVBC could have a significant impact in supporting BCSs throughout the cancer continuum:

> *There were a lot of doubts, fevers, discomforts, disquietudes...there still are...and if I had this type of resource, I could have used it instead of googling or looking into patient groups, for some kind of answer...Because you do feel the need to go to forums and ask, does anyone feel like this? What have you done? And if there were a platform like this, the type of support provided would be different, more credible and appropriate.* [BCS11]

Similar to BCS11, most participants valued the possibility of accessing "trustworthy" (BCS6) self-care information provided "anytime, anywhere" (BCS7) via iNNOVBC. The interviewed BCSs had searched on the web for information on treatments' adverse events, practical strategies, and emotional challenges they were faced with, but their searches were lengthy and required them to assess the quality of information they were presented with. Both BCSs and MHPs considered that having "easy access to evidence-based ready to use psycho-educational

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content" (MHP10) could help survivors better manage their condition and problems.

According to the participants, the survivorship trajectory is characterized by many biopsychosocial challenges that deeply impact survivors' well-being. Several participants described the pervasive and long-lasting impact that treatment's adverse events or sequelae had on their physical and mental health, underlining the importance of receiving accurate and comprehensive information about it. Effects such as alopecia, onycholysis, pain, menopausal symptoms, fatigue, cardiotoxicity, lymphedema, osteoporosis, infertility, and memory loss were often discussed by interviewees as key information points to address in a nuanced and dynamic manner. During the course of treatment, various survivors received flyers and booklets about treatments' adverse effects, but most complained about their unappealing design and low intelligibility and focus on the active treatment stage, therefore failing to provide a continuous perspective on its management after treatment completion. The fact that iNNOVBC approached many of those themes "in a chronological way" (BCS1) was appreciated by various BCSs because it enabled them to prepare and cope with it in the long term.

The impact of cancer and its treatments on survivors' emotional health was another matter of concern. Both MHPs and BCSs underlined the importance of providing psychological support to BCSs throughout the cancer continuum, particularly during the transition from active treatment to follow-up care:

> *Getting psychological support is very important at all stages...at the diagnosis, during treatment and after treatment...People are not aware of this, they tend to say "Ah, that's over now!" and expect us to return to whom we used to be...But that's not possible...you still need support. This feels almost like...a post-traumatic situation...and by saying that it's almost like they're taking away your legitimacy to feel the pain, so it can feel very isolating to cross that path.* [BCS2]

> *It's an abrupt shift from being extremely cared for, from having that unconditional support during chemo to stopped being cared for...As soon as your hair starts to grow, everyone assumes that everything is fine. But it's not. I will never be able to say again that I'm fine. I'm not healed, I'm full of fears, anxiety, and sorrow. So...it was extremely hard for me to deal with that because I felt abandoned...people had pulled the rug out from under me.* [BCS7]

Similar to BCS2 and BCS7, various participants discussed the experience of feeling in distress and unsupported after finishing the primary treatment. Problems such as "dealing with low mood" (BCS11), "feeling anxious all the time" (BCS2), "fearing that cancer had returned" (BCS8), or fear of "dying" (BCS9) were common, often impacting BCSs' sleep and interpersonal relationships. Difficulties in adapting to a "scarred body image" (BCS5) were also prevalent in interviewees' narratives and various participants reported on the profound impact it had on their self-esteem, sexuality, and intimate relationships:

> *Losing my breast shook my self-esteem, it shook everything...it's hard to live without it, I'm a woman. I...I'm incapable of being naked in front of my husband. I feel embarrassed...I'm not able to hug him anymore. I'm afraid he feels that it's not my breast, that it isn't real. I sleep...without the prosthesis, but I always try to sleep with my back to him and when I realize I'm facing him, I turn over right away...I'm afraid that if he sees me, he might lose his interest in me, that he stops desiring me. ...We still have sex but it's not the same. I'm always afraid that he touches where he's not supposed to...I do not lean against him...I avoid his touch. It's difficult and...I know that there are women that deal with this well, but I...I do not.* [BCS9]

Similar to BCS9, various participants talked about not being able to adapt to their changed self, feeling like a fake version of themselves, incapable of restoring lost intimacies, and isolated by the "tabu cancer had become" (BCS2) in their inner circles. Coping with such problems was highly demanding, and many struggled to discuss such topics with friends, family, and professionals owing to their sensitive nature. Thus, many interviewees appreciated the "assertiveness of the themes" (BCS6) explored at iNNOVBC and the possibility of receiving professional support at a distance or without face-to-face contact:

> *It's important to have support but it's uncomfortable to talk about these issues face-to-face...Maybe through this platform, it could be easier...I believe I could feel more comfortable to ask some questions if I knew this was anonymous, if I didn't have to identify me...I believe it would be much easier to approach intimate topics via chat or e-mail than in-person.* [BCS3]

Participants such as BCS3 appreciated the fact that iNNOVBC allowed direct communication between MHPs and BCSs. Both professionals and survivors considered having chat, email, and videoconference communication alternatives to be helpful. By offering synchronous and asynchronous communication channels associated with different degrees of exposure, participants considered that iNNOVBC "could promote survivors' self-disclosure" (MHP6) and facilitate the "timely discussion" (MHP4) of sensitive topics, ultimately, "benefiting the therapeutic process" (MHP12). They also saw advantages in the possibility of automatically sending psychoeducational content and scheduling and notifying BCSs about tasks and questionnaires to be completed along the implementation of the program:

> *The functionalities are well thought and the fact that everything is integrated is awesome...this combines a modular treatment approach with videoconference and chat...it allows you to take notes...and the instruments are embedded...That's awesome and exactly what I need in my practice!* [MHP5]

The fact that iNNOVBC combines evidence-based content with communication, monitoring, and documentation features was praised by various professionals. The interviewed MHPs considered that it could reduce the time and effort they need to

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invest in finding, preparing, and assessing materials, enabling them to work more efficiently. iNNOVBC was also considered useful to promote "continuity of the relationship established between the therapist and the client" (MHP7) beyond appointments. Most importantly, by doing so, iNNOVBC could configure a point-of-need service that could support BCSs along the survivorship trajectory. Nevertheless, it is important to say that iNNOVBC's usefulness could be compromised by its "lack of intuitiveness" (MHP9).

Usability

Both the quantitative and qualitative data collected during the usability tests and interviews identified design and functionality issues that could be improved to enable more effective and efficient use of iNNOVBC and increase users' satisfaction with the program. In general, participants' effectiveness, as measured by completion rates, was high in both groups (range 69.2-100; Tables 2 and 3), but the System Usability Scale median score was 65 (IQR 35) for BCSs and 47.5 (IQR 25) for MHPs, which may be considered below average in terms of usability. These results are aligned with the usability issues identified during the usability tests.

Table 2. MHP[a] laboratory-based usability test results.

Task groups and tasks	Task performance, %	Error, mean (SD)	Assistances, mean (SD)
A: Log-in			
1. Log in to the iNNOVBC[b]	100	0.2 (0.4)	0.1 (0.3)
B: Notifications			
2. Check notifications	84.6	0.8 (1.2)	0.4 (0.6)
3. Comply with the notifications' instructions by accessing the patient's file	100	2.3 (2.6)	0.7 (0.8)
C: Treatment prescription			
4. Click on the patient's link or search for the patient	100	3.2 (5.1)	0.7 (1.1)
5. Check the modules prescribed to the patient	100	1.1 (1.2)	0.2 (0.4)
6. Assign the sleep module to the patient	100	0.6 (1.1)	0.1 (0.3)
7. Send a notification to the patient	69.2	0.4 (0.7)	0.3 (0.8)
8. Save the previous procedure	100	0.1 (0.3)	0.2 (0.4)
9. Check the available clinical trials	100	3.2 (6.6)	1 (1.3)
10. Prescribe iNNOVBC to the patient and schedule the onset of the treatment	100	0.6 (1.3)	0.4 (1.1)
11. Save the previous procedure	100	0 (0)	0.1 (0.3)
D: Treatment progress assessment			
12. Send feedback to the patient about the homework assignment	100	3.8 (5.1)	1.1 (1.2)
13. Access the patient's file	100	0.7 (1.4)	0.4 (0.7)
14. Click on questionnaires	100	3.8 (3.0)	1.4 (1.4)
15. Check the questionnaires available to prescribe	100	1.6 (3.4)	0.4 (0.8)
16. Assign the weekly questionnaire to the patient	100	1.5 (2.4)	0.6 (0.8)
17. Save the previous procedure	100	0 (0)	0 (0)
E: Conversations			
18. Write an email to the patient	100	1.6 (2.1)	0.5 (0.6)
19. Send the email	100	0 (0)	0 (0)
20. Start a chat conversation with the patient	100	1.5 (2.8)	0 (0)
21. Start a videoconference appointment with the patient	100	0.1 (0.3)	0.1 (0.4)
22. Update the patient's clinical diary	100	4.4 (5.2)	0.8 (1.2)
23. Schedule the next appointment and set an alarm	100	0.6 (0.8)	0.1 (0.3)

[a]MHP: mental health professional.

[b]iNNOVBC: iNNOV Breast Cancer.

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Table 3. BCS[a] laboratory-based usability test results.

Task groups and tasks	Task performance, %	Error, mean (SD)	Assistances, mean (SD)
A: Log-in			
1. Log in to iNNOVBC[b]	100	0.6 (0.8)	0.5 (0.7)
B: Notifications			
2. Check notifications	81.8	1.3 (1.6)	1.7 (2.5)
3. Read the therapist's message	100	0.5 (1.2)	0.4 (0.8)
C: Treatment content management			
4. Access treatment modules	100	1.9 (2.6)	2.4 (1.8)
5. Access the relaxation module	100	0 (0)	0.4 (1.1)
6. Open the deep muscle relaxation page and expand the *how to practice* text	100	2 (1.9)	2 (1.5)
7. Play the recorded relaxation session	100	0.5 (0.9)	0.7 (1.4)
8. Download the relaxation session	100	0.1 (0.3)	0.4 (1.1)
9. Print the current page	100	0.2 (0.4)	0.6 (0.9)
D: Worksheet completion			
10. Return to the modules	100	0.5 (1.4)	0.9 (2.3)
11. Access page 8 of the anxiety module	100	0.1 (0.3)	0.1 (0.3)
12. Complete and save anxiety ladder exercise	100	0.3 (0.6)	0.4 (0.9)
13. Access the sleep diary	100	0 (0)	0 (0)
14. Complete the sleep diary	100	0.5 (1.2)	1.3 (2.5)
E: Communicating with therapists			
15. Send an email to the therapist	100	0.5 (0.8)	0.6 (0.9)
16. Start a chat conversation with the therapist	100	1.1 (1.3)	0.2 (0.6)
17. Start a videoconference appointment with the therapist	100	0 (0)	0.5 (1.2)
F: Scheduling tasks			
18. Schedule a new task and set an alarm	100	0.9 (0.7)	0.8 (1.7)

[a]BCS: breast cancer survivor.

[b]iNNOVBC: iNNOV Breast Cancer.

According to the classification by Zahabi et al [62], the issues identified during the usability tests (Multimedia Appendix 7) were mostly related to inefficient interaction (12/43, 28%), ineffective information presentation (9/43, 21%), cognitive overload (7/43, 16%), ineffective use of language (6/43, 14%), and lack of naturalness (ie, lack of familiarity or matching between users' usual workflow and the system; 6/43, 14%). Issues related to consistency (5/43, 12%), feedback (3/43, 7%), customizability and flexibility (3/43, 7%), and error prevention (2/43, 5%) were also identified, although less frequently.

Throughout the usability tests and posttest interviews, participants commented on the importance of making iNNOVBC's information architecture clearer to facilitate navigation. Various participants verbalized difficulties in understanding how the information and features made available on the platform were hierarchized and could be managed, thus requiring a simplification of navigation paths:

> *I was always questioning what was for the therapist and what was for the patient...What resources were available to me or them...What was available to prescribe and what was already prescribed...So...I think this should be made clearer.* [MHP6]

> *I was a little bit lost because I couldn't find the way...If it were like Pinterest...or facetime, I believe it would be more accessible...[because] All the little windows appear right away, and the images are clear and appealing to me.* [BCS4]

Similar to BCS4, various participants considered changes to iNNOVBC's user interface could be performed. Some participants believed the program could be redesigned to display "a unique dashboard with all key features available at login" (BCS8), whereas other interviewees suggested that a tour providing an overview of the platform or adding labels and preview options to most used functionalities would suffice. Furthermore, the use of familiar and interactive design was also appointed as a strategy that could facilitate navigation:

> *I found it a little monotonous, everything looked the same...and that confused me. Maybe using more*

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> *colour and highlighting some parts...it would be easier to understand how the materials are organized....* [BCS6]

Similar to BCS6, various participants expected to find a more appealing and dynamic color scheme, as well as greater use of icons, images, and shortcuts. Despite the perceived adequacy of the content materials, the presentation of long text, sometimes displayed along various binders and features, was considered problematic. Various participants had to do a strenuous effort to screen through the available information and complete the usability test tasks. As a result, participants often became frustrated and some verbalized feeling discouraged to use the program:

> *I find the anxiety module content very clear, easy to read and very interesting for anxiety sufferers...but the program, in terms of presentation, requires some improvements because sometimes the information displayed is too much.* [BCS2]

> *Not only from the therapists' perspective but also from the patients' there are things that should be simplified so that they do not experience the same frustration I felt because they would give up on using the program...Instead of making it easier, I am afraid they would have to press so many buttons, that they would not understand what is expected from them, and they would give up...ending up not sending anything.* [MHP3]

Corroborating MHP3, various participants mentioned that simplifications to the provided materials should be performed. Participants suggested organizing the information more concisely, in accordance with single themes, providing hyperlinks to additional information whenever necessary. Participant BCSs also underlined the importance of simplifying the exercise worksheets owing to difficulties in handling tables (eg, sleep and symptom diaries), suggesting that information should be entered using plain textboxes:

> *It should have an option to enter the date and then the system would label it as register 1, 2, 3, etc...automatically. This shouldn't be a table, it should be a simple field to complete and then the psychologists would see the table. If it's important to the patient to see the progress, a different graphical approach should be used...considering that there are a lot of items to be filled in the platform, I believe having a summary, and overview...that helped me realize my progress...maybe presented as a graph, not only in writing...and something beautiful to see...would make me feel, you know?...Wow, I managed to get here today...As children have at school, the green, yellow, and red stars...Maybe I'm being childish...but for those who are at this stage...Positive reinforcement is needed....* [BCS11]

Similar to BCS11, the MHPs participating in the study also recommended the integration of data visualization dashboards into the program. Professionals considered the inclusion of a "simple dashboard providing digested information about specific scores, or cut-offs being exceeded or any tasks or questionnaires pending" (MHP7) would facilitate the handling of the program and BCSs' treatment progress follow-up. The inclusion of gamification principles was also mentioned as interesting by a few MHPs, owing to its potential of promoting engagement to treatment in BCSs.

The use of unfamiliar terminology (eg, users/*utilizadores*, treatment modules/*módulos de tratamento*, and worksheets/*Fichas de trabalho*) and a perceived lack of integration between some sections of the platform was also a matter of concern. Various participants struggled to grasp iNNOVBC's affordances because the designations used were unfamiliar to them. Moreover, various participants verbalized difficulties in learning and remembering how to navigate from the user hub or the treatment modules section to the conversations section and vice versa. During this process, errors were recurrent, and many participants opted for a trial and error or a *go to landing page* strategy to complete the proposed tasks. These difficulties hindered not only participants' effectiveness and efficiency but also their satisfaction with the program:

> *The terms used weren't completely obvious...It was hard for me to understand how to find the patient, how to consult the things that she had performed, the tasks I had assigned to her...It confused me because those were not the terms I use in my practice...I believe something closer to what I use daily, closer to the platforms we have there [at the hospital] would be easier...like a list of patients...clinical file...prescriptions...results....* [MHP12]

> *Imagine I am reading something, and I am not understanding it well or I want to tell what is happening to me to the therapist...Just the fact that I must go to another page and look for it [conversations section] creates a huge mess because I do not know where to go and when I get there I do not remember anymore where I was at....* [BCS7]

> *...I would have to have a paper beside me to write down the doubts that are arising, or I would have to use two screens...Minimizing one to get to the other...It would make some sense, yes, to have it [the chat] always available.* [BCS2]

As mentioned by BCS2, various participants considered that some features, such as the conversation's menu, should be always on display. Survivors considered it to be important to express doubts, concerns, and emotions while reading the treatment modules or performing the exercises. Therapists expressed the need to easily provide feedback to their clients and consult communication logs while executing other tasks. In addition, some participants suggested that a customizable toolbar should be made permanently available to facilitate the use of the platform.

The importance of being able to customize the program and use it in a flexible way was reiterated by various participants. The possibility of selecting the treatment modules to work with, how the information is conveyed (eg, audio, video, and text), and between alternative versions of the same material (eg, male and female relaxation audios) was considered important to make the program more inclusive and engaging:

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> *Audios and videos...If the person doesn't want to read or has a lot of difficulties in understanding, that could help...and, the way I see it, this shouldn't be an obligation, but an app that is available to help me and to be used according to my needs. For example, the relaxation...I am working on my sleep isn't it, controlling my sleep and then I could access the other menus like...Now I am going to relax for a while, and then have the exercise 1, 2 and 3 and...ok, today I'm going to use this one....* [BCS8]

> *Maybe I like having more control...But I believe it's important to me to be able to decide which module a specific patient should be working with in a particular week...Depending on what I think that makes more sense to the person at that specific time...or to be the client herself deciding what makes more sense...I believe that for this to be accepted in the future, professionals must have the possibility of adding information or questionnaires themselves. I think that's important, to make them feel this is a valuable tool and not to reject it.* [MHP13]

Similar to MHP13, various participants mentioned the intention to use the program as an *à la cart* platform or self-care toolbox, where different content and strategies could be prescribed simultaneously or used as needed. This was also clear from the participants' behavior during the usability tests, where sometimes errors were committed because participants assumed that modules (eg, the anxiety, sleep, and relaxation modules) could be used interchangeably and not sequentially. This discrepancy between the program's original concept and how participants appropriated the program or intended to use it was emphasized during the field trials, impacting participants' perception of iNNOVBC's feasibility.

Feasibility

Many participants reported that integrating iNNOVBC into their routine, as prescribed, would be feasible but challenging. MHPs worried about the implementation of the program at their workplaces owing to interoperability as well as practical and attitudinal limitations, whereas BCSs anticipated difficulties in "finding the time to fit the program into the day-to-day life" (BCS1):

> *Considering the hustle of professional life, I believe it wouldn't be easy for me to comply with everything that is asked...to enter the site and make daily registrations because I would have to be available to complete several steps and to pay close attention to it. Nowadays, people want simple, fast, and short tasks...and as is, I am not sure I would be able to do it straightforwardly.* [BCS6]

> *It is important to me to have some flexibility...I don't want to make this another chore that I have to do. I think it would make me even more stressed because I would be worried about complying...I don't want it to be another obligation.* [BCS3]

As mentioned by BCS3 and BCS6, various interviewees underlined the role that flexibility, accessibility, and ease of use could play in iNNOVBC's uptake. During the field trials, various participants used iNNOVBC on their smartphones, owing to its portability and usability. A participant who was retired justified that she did not "use computers in a long time, being more acquainted with mobile devices" (BCS4). Another participant who was on sick leave mentioned not using computers regularly, because she did not usually carry it with her anymore and some "movements, like pulling the plug or pressing the mouse buttons, still hurt due to hormonal treatment" (BCS9). Another working participant revealed being receptive to use the web version of the program but showed some resistance in spending additional hours in front of the computer while at home (BCS10). However, as iNNOVBC runs on a responsive web-based platform, some materials did not perfectly adapt to their mobile devices and some exercises such as the relaxation audios were interrupted by notifications, thereby compromising the delivery of the intervention. Thus, some participants reported it would "be easier to have it in app format" (BCS11), suggesting that this is a more convenient and accessible format for BCSs.

Another important aspect discussed by BCSs as possibly facilitating adherence to the program concerned having a direct communication channel with psychologists:

> *Having a psychologist on the other side of the screen is particularly important...Sometimes you need to listen to someone assertive to be able to keep going...If this were like an online helpline like SNS24 [a telephone and web-based service of the Portuguese National Health Service that provides support to citizens when they need advice with acute, nonemergent health complaints] but for psychological issues, I would use it often. If I saw it was reliable and that it helped me, I believe I would use it several times, because I missed that support.* [BCS7]

> *Having timely feedback on what to expect or about what is considered normal and not having to wait for the next appointment could be very important to better manage the emotional impact of cancer, making me interested in using this.* [BCS10]

The possibility of posing doubts; discussing difficulties associated with the treatment, follow-up, and discharge processes; and receiving professional feedback from psychologists was valued by most BCSs. Interestingly, survivors seemed to conceptualize iNNOVBC not necessarily as a structured psychotherapeutic approach but as an on-demand tangible supportive tool that could provide them with access to tailored content and psychological support timely delivered according to the specific moment of the survivorship trajectory they were at. According to participants, both information and supportive care needs change significantly along the survivorship continuum and having access to such a tool could secure them that they were "still being taken care of" (BCS1) during the follow-up stage.

However, to fulfill its full supportive role, besides performing some simplifications to the program, most participants considered that some sort of training should be provided to both MHPs and BCSs. Participants considered that having access to training material in graphical, written, or video format would

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facilitate the use of the program. In addition, some MHPs mentioned that considering the novelty of the programs' approach, the provision of training sessions and on-job training would be advisable not only to present the platform but also to introduce therapists to the program's rationale and components:

> *It is important to know more about the program, how it was created and that it really works. It would provide us with more confidence in using the program if we knew we would not be wasting our clients' time and money.* [MHP9]

> *I believe training should be an extended version of what we have done here today [usability test], with practical situations and instructions, so we can clarify our doubts...like a workshop session...and then having someone there to help us during the first weeks....* [MHP12]

The positive impact of having dedicated professionals, conceivably digital navigators, supporting the implementation of the program was also discussed by BCSs. Some participants considered it would be important to have an appointed professional to introduce them to iNNOVBC's content, structure, and features, at their cancer centers. Such support could help them in overcoming usability issues and attitudinal barriers toward the program and, ultimately, facilitating adoption:

> *After being admitted or having a first appointment with the doctor, he could say "now you are going to meet a colleague of mine, that will show you how to use a tool that can help you deal with your situation" and then the designated colleague, that has to be an appealing person, possibly a psychologist to better know how to convey the information to the person, would explain the purpose of the program, show how the program works and even trial it with the patient...in the beginning, they might think "this is boring", but If people are properly introduced [to the program], later, when they are feeling more anxious, they might remember "Ah, I have that app that can help me 24/7" and they would value the support that is provided in here.* [BCS8]

iNNOVBC was regarded as part of a comprehensive portfolio of services to be provided by cancer centers to BCSs, to which they would have to be informed about, introduced to, and properly referred to, to be able to use it at its full potential. Similar to BCS8, some MHPs thought that oncologists could play a significant role in facilitating iNNOVBC dissemination and uptake in cancer settings. As physicians "are the ones orchestrating treatment" (MHP5), they were viewed by participants as important gatekeepers of programs such as iNNOVBC. However, some MHPs mentioned that oncologists could not necessarily be "receptive to prescribe it, due to their biomedical approach...and lack of involvement in the delivery of psychosocial interventions" (MHP2). Thus, for iNNOVBC to become part of survivors' "adjuvant supportive care" (MHP1) and for it to be properly integrated into clinical settings, some MHPs considered that training should be extended to other professional groups, such as nurses, physicians, and managers working within oncology. To be successfully implemented, iNNOVBC would have to be recognized as a valuable service to be provided by the several actors playing in the cancer setting.

Discussion

Principal Findings

In this study, iNNOVBC—a guided, internet-delivered, individually tailored, ACT-influenced CBT intervention aiming at treating mild to moderate anxiety and depression as well as improving fatigue, insomnia, sexual dysfunction, and HRQoL in BCSs—was developed with a user-centered design approach [37] and explored concerning its usefulness, usability, and feasibility.

Overall, participants considered iNNOVBC highly useful, with most interviewees reporting on the pertinence of its scope, digital format, content, and features. Consistent with the literature [63], participants reported on the high prevalence of physical, emotional, practical, and information unmet care needs experienced by BCSs and considered that iNNOVBC could help bridge the supportive care gap experienced by BCSs across the survivorship trajectory. Similar to previous research [64-66], survivors valued having access to ubiquitous evidence-based self-care information that was organized in accordance with the cancer continuum, considering that it could help them better manage and cope with their condition and problems.

Another important aspect contributing to participants' perception of the usefulness of iNNOVBC concerned the multifeatured and integrated nature of the program. Participants appreciated the fact that iNNOVBC combined psychoeducation, communication, documentation, and automatized scheduling and notification features, allowing a more efficient and comprehensive assessment and follow-up of BCSs. Furthermore, the possibility of MHPs and BCSs to communicate synchronously and asynchronously (eg, via chat, email, and videoconference) through the platform, as well as in accordance with different degrees of personal exposure, was valued by various participants. Some participants mentioned that using chat or email could facilitate self-disclosure in BCSs and promote the timely discussion of sensitive topics often avoided, thus having a positive impact on the established therapeutic alliance and process. Nevertheless, few studies have addressed chat-based internet interventions targeting cancer survivors. Previous research has focused on chat groups for patients with prostate cancer [67] or adolescents treated for cancer and reported mixed results. Thus, it is necessary to conduct further research on one-to-one chat-based programs to evaluate the role chat sessions could have in survivors' treatment progress and engagement in digital supportive care.

Despite participants' perceived usefulness of iNNOVBC, both BCSs and MHPs identified aspects hindering their experience while using the program and changes to be performed to achieve better effectiveness, efficiency, and satisfaction with iNNOVBC. These included refining the program's aesthetics by using a minimalist and recognizable design; improving interaction design by making the navigation within the program more consistent and constrained; decreasing the cognitive overload experienced by participants by using terminology tailored to

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each context of use and balancing the amount of information displayed; increasing the feedback provided to users, so that they are continuously informed about the impact of their work within the program; and diversifying the media used for intervention delivery by developing smartphone and tablet versions of the program. These findings echo many of the limitations and development requirements gathered in previous research aimed at developing digital programs for supporting cancer survivors [64,65,68-73].

In particular, BCSs and MHPs participating in this study requested the simplification of navigation paths, suggesting that the use of an opening dashboard would facilitate the understanding and handling of iNNOVBC. Adding labels and preview options to the most used functionalities was also considered important to increase the discoverability of the program. Likewise, the use of a more dynamic color scheme and greater use of icons, images, and shortcuts were appointed as necessary to facilitate the recognition of the program's affordances and facilitate its use. Previous research has yielded comparable results [64,65,69-71], suggesting that single-page websites or apps that enable properly labeled interactions and make use of different color depths, familiar icons, and images are more usable and acceptable to cancer survivors.

Moreover, participants underlined the importance of balancing and diversifying the information that is displayed at each given time, as well as simplifying data entry tools. According to participants, information should be grouped more concisely (eg, short modules addressing a single theme), delivered using various media (eg, web, smartphone, and tablet), and displayed in different formats (eg, text, audio, and video) and versions (eg, female and male audio clips) and should allow entering data using plain textboxes instead of tables to make the program more inclusive and easier to use. Complementarily, the integration of data visualization dashboards providing digested information on completed or to-be-completed tasks and questionnaires, its scores, and cutoff points being exceeded or achieved was appointed by interviewees as relevant. Participants anticipated that such a strategy could facilitate the handling of the program and the assessment of BCSs' treatment progress, promoting their engagement with iNNOVBC. Previous research corroborate these findings [71-73]. In a previous study by Igelström et al [71], the importance of delivering content in different formats and adding a graphical display of self-reports was emphasized by participant survivors. In another study by Wagner et al [72], BCSs stressed the importance of developing a *my progress page* to display didactic content and tools that had been completed and chart anxiety scores to facilitate tracking of progress. Nevertheless, and according to Kuijpers et al [74], although professionals might be primarily interested in dashboards indicating a worsening of symptoms to help patients reduce symptom burden, survivors seem to be interested in monitoring changes in their symptom experience and functional health, preferring to see both worsened and improved scores depicted in such visualizations. These results underline the importance of tailoring DMH programs to the profile and unique preferences of each user to better address their concerns and needs.

The option to tailor iNNOVBC along the cancer continuum was a salient development requirement identified during this study. Being conceived as an individually tailored program, iNNOVBC permits some degree of tailoring, namely, in what concerns the treatment modules. However, participants considered further layout and content customization should be allowed so that the program could adapt to the idiosyncrasies of each of the survivorship trajectory stages. Participants considered that comprehensive support should be provided along this continuum, not compartmentalizing survivors' needs but shaping and transforming the program according to its evolution. Similar to a rhapsody, iNNOVBC should adopt "an episodic yet integrated, free-flowing structure, featuring a range of highly contrasted moods, colour, and tonality" [75], to be used freely and flexibly as needed. This finding aligns with previous research [66,72,76,77] and emphasizes the importance of building flexibility into digital programs targeting cancer survivors, not only in terms of tailoring but also in terms of frequency and timing of use, to ensure its uptake by target users.

However, such an understanding of the program contrasts with its original concept and theoretical grounding. Although participant survivors mentioned the intention to use the program as an *à la cart* platform or self-care toolbox, where different content and strategies could be prescribed simultaneously or used as needed, and not in a prescriptive manner, evidence suggests that the implementation of structured approaches, theoretically grounded and validated to specific contexts of use, best serves survivors [13,14]. This discrepancy adds to the technical, usability, funding, attitudinal, and training limitations identified by participants and the literature [51] as potentially hindering successful implementation of iNNOVBC and underlines the need to further assess and refine it in clinical contexts before scaling up the program. Thus, iNNOVBC will soon be pilot-tested in cancer settings [78] not only to assess its preliminary efficacy but also to further assess its feasibility and gather requirements for the design of a patient-centric service that fits into BCSs' lives, professionals' evidence-based practices, and cancer centers' workflows. After piloting and further refining iNNOVBC, the program will be tested for its efficacy and cost-effectiveness using a multicenter, randomized, waiting list, controlled design [78]. The results from this parent study will determine whether iNNOVBC should be transferred to routine care.

Strengths and Limitations

This study presents various strengths and limitations. Strengths of this study include the adoption of a user-centered design approach combining mixed methods (eg, surveys, in-depth interviews, usability tests, and field trials) that were used to explore the usefulness, usability, and preliminary feasibility of iNNOVBC in a comprehensive manner. Furthermore, the study was conducted by an interdisciplinary team and involved iNNOVBC's primary and secondary end users, that is, BCSs and MHPs, benefiting from complementary input by these stakeholders. In addition, participants have been purposefully sampled for diversity in age, academic degree, and digital technology proficiency. Limitations include a small sample size and minimal diversity in participants' educational level and experience in using DMH programs. Moreover, participant

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BCSs were younger in the current sample than the general Portuguese BCS population [79], and participant survivors were not screened for mild to moderate anxiety or depression. This aspect may have prevented the identification of usability issues associated with older age or psychological morbidity, thereby limiting the overall generalizability of our results. Nevertheless, there is some consensus that usability tests may include a minimum of 5 participants per iteration and that approximately 80% of usability issues are discovered with as few as 4-6 participants [80]. Furthermore, many of the results obtained in this study align with previous research, supporting its ecological validity. Future research [78], aiming at pilot-testing and further assessing iNNOVBC's feasibility, efficacy, and cost-effectiveness, should include older and less technically adept participants to complement the findings of this study.

Implications for the Design and Implementation of DMH Programs in Cancer Settings

The results of this study hold important implications for the further development and implementation of programs such as iNNOVBC. First, DMH programs targeting cancer survivors could benefit from the early involvement of its primary, secondary, and tertiary end users in the development process. By involving survivors, health care professionals, and managers in codevelopment activities (eg, surveys, in-depth interviews or focus groups, usability tests, and field trials), interventions could be designed to address stakeholders' real needs and development could better align with their practices and contexts, thereby increasing the odds of successful implementation. Second, involving interdisciplinary teams in the development process is key to ensure that comprehensive solutions are developed and design caveats are timely anticipated, identified, and refined, thus not compromising the usefulness, usability, and feasibility of such programs. Third, DMH interventions must be conceived as supportive point-of-need services capable of adjuvating and extending cancer care delivery. Programs must adopt a flexible yet integrated structure capable of being continuously tailored to end users' changing needs, evolving along the cancer continuum. In this context, transdiagnostic programs might be particularly useful in fulfilling this requirement. Finally, implementation research must be conducted to determine the effectiveness of developed programs and identify service delivery bottlenecks (eg, lack of training) to which fast-track solutions (eg, digital navigators) must be developed and tested.

Conclusions

This study explored the usefulness, usability, and preliminary feasibility of iNNOVBC, and its results suggest that DMH programs, such as iNNOVBC, are considered useful by both BCSs and MHPs, thus configuring a promising point-of-need solution to bridge the supportive care gap experienced by BCSs across the survivorship trajectory. However, to fulfill its full supportive role, such programs must be comprehensive, highly usable, and tailorable and adopt a flexible yet integrated structure capable of evolving in accordance with survivors' changing needs along the cancer continuum.

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Authors' Contributions

CMS, FN, EW, RS, and GA conceptualized and designed this study. CMS carried out the acquisition of data. George Vlaescu developed and configured iTerapi to serve the purposes of iNNOV Breast Cancer, providing important technical and intellectual support for the development of the program. CMS wrote the manuscript; CMS and FN analyzed and interpreted the data. FN, EW, RS, and GA revised the paper for important intellectual content.

Conflicts of Interest

None declared.

Multimedia Appendix 1
Study intervention components.
[DOCX File , 25 KB - cancer_v8i1e33550_app1.docx]

Multimedia Appendix 2
Breast cancer survivors' and mental health professionals' interview scripts.

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[DOCX File , 24 KB - cancer_v8i1e33550_app2.docx]

Multimedia Appendix 3
Breast cancer survivors' and mental health professionals' iNNOV Breast Cancer usability test protocol and observation tables.
[DOCX File , 35 KB - cancer_v8i1e33550_app3.docx]

Multimedia Appendix 4
Breast cancer survivors' and mental health professionals' debriefing interview scripts.
[DOCX File , 18 KB - cancer_v8i1e33550_app4.docx]

Multimedia Appendix 5
Field trial protocol.
[DOCX File , 14 KB - cancer_v8i1e33550_app5.docx]

Multimedia Appendix 6
Usefulness and feasibility questionnaire.
[DOCX File , 19 KB - cancer_v8i1e33550_app6.docx]

Multimedia Appendix 7
Identified usability issues.
[DOCX File , 32 KB - cancer_v8i1e33550_app7.docx]

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Abbreviations

ACT: acceptance and commitment therapy
BCS: breast cancer survivor
CBT: cognitive behavioral therapy
DMH: digital mental health
HRQoL: health-related quality of life
iNNOVBC: iNNOV Breast Cancer
MHP: mental health professional

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Original Paper

Clinicians' Perceptions of the Benefits and Challenges of Teleoncology as Experienced Through the COVID-19 Pandemic: Qualitative Study

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Abstract

Background: COVID-19 thrust both patients and clinicians to use telemedicine in place of traditional in-person visits. Prepandemic, limited research had examined clinician-patient communication in telemedicine visits. The shift to telemedicine in oncology, or teleoncology, has placed attention on how the technology can be utilized to provide care for patients with cancer.

Objective: Our objective was to describe oncology clinicians' experiences with teleoncology and to uncover its benefits and challenges during the first 10 months of the COVID-19 pandemic.

Methods: In-depth, semistructured qualitative interviews were conducted with oncology clinicians. Using an inductive, thematic approach, the most prevalent themes were identified.

Results: In total, 21 interviews with oncology clinicians revealed the following themes: benefits of teleoncology, such as (1) reducing patients' travel time and expenses, (2) limiting COVID-19 exposure, and (3) enabling clinicians to "see" a patients' lifestyle and environment, and challenges, such as (1) technological connection difficulties, (2) inability to physically examine patients, and (3) patients' frustration related to clinicians being late to teleoncology appointments.

Conclusions: Teleoncology has many benefits and is well suited for specific types of appointments. Challenges could be addressed through improved communication when scheduling appointments to make patients aware about what to expect. Ensuring patients have the proper technology to participate in teleoncology and an understanding about how it functions are necessary.

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KEYWORDS

teleoncology; telemedicine; qualitative; COVID-19; telehealth; cancer care; cancer; oncology; digital health; pandemic

Introduction

Telemedicine, defined by the Institute of Medicine as the use of electronic information and communications technologies to provide and support health care when distance separates the participants [1], was not often utilized in cancer care prior to COVID-19 [2-4]. Although advocates of telemedicine called for improved access to the technology before COVID-19 [5-7], the pandemic forced health systems to rapidly adapt. For instance, a study evaluating claims data found that telemedicine utilization for office visits and outpatient care was 78 times higher in April 2020 than in February 2020 among various

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diseases, including cancer [8]. Telemedicine is enabled by over 90% of adults in the U.S. using the internet, although only 77% have broadband internet service at home [9]. The surge was a result of loosening regulations, which allowed for insurance coverage and reimbursements for telemedicine visits [10]. In 2021, the American Society of Clinical Oncology (ASCO) published standards and practice recommendations to ensure that clinicians effectively use telemedicine with their patients now and in the future [11]. However, the review summarized previous telemedicine studies and focused on situations when it was most appropriate to deliver care rather than how patients and clinicians interact with one another using the technology.

In cancer care, effective clinician-patient communication is particularly important because it impacts patients' psychosocial outcomes and quality of life [12]. Prepandemic, limited research had examined clinician-patient communication in telemedicine visits. In a study consisting of interviews with oncology professionals (eg, physicians, physician assistants, and nurse practitioners) about using telemedicine, Heyer et al [13] discovered that clinicians were concerned about whether they could effectively build rapport and provide patients with the support necessary to nurture clinician-patient relationships that are integral to quality care. The study was conducted between October 2019 and March 2020, immediately preceding the COVID-19 pandemic [13]. Questions remain about whether these perceptions persisted during the pandemic, as telemedicine became entrenched in the health care delivery experience. A recent paper that surveyed both patients and cancer clinicians during the pandemic found that patients are more enthusiastic about using telemedicine than clinicians, with a greater number of responses stating that clinicians prefer in-person visits [14].

We use the term "teleoncology" in this study to refer to visits between cancer patients and clinicians conducted over videoconferencing applications, such as Zoom (Zoom Video Communications). The rapid shift during the first few months of the COVID-19 pandemic to teleoncology [15] has provided an unprecedented opportunity to understand oncology clinicians' experiences with the technology. Research has assessed the patient perspective in cancer care, finding that patients experience technical difficulties [16] but are also largely satisfied with the encounter [17]. Thus, the purpose of this qualitative study was to describe oncology clinicians' perceptions of teleoncology and to identify its benefits and challenges during the first 10 months of the COVID-19 pandemic.

Methods

Study Design

We conducted an in-depth qualitative study at the University of Florida Health Cancer Center (UFHCC). The UFHCC is a 192-bed hospital serving North Central Florida, specializing in 14 cancers, such as blood cancer, lymphoma, breast cancer, and head and neck cancer. The cancer center serves surrounding rural counties, which make up 20% of the patient population. The University of Florida Institutional Review Board (202000243) approved the study, and all participants consented to participate before interviews began.

Participants and Recruitment

Inclusion criteria consisted of participants being clinicians (oncologists, nurse practitioners, or physician assistants) who provided care to individuals with a cancer diagnosis and were willing to participate in an interview. We sent an email and 1 reminder email to all medical and radiation oncology clinicians at our cancer center with a description of the study and a link to an online screening questionnaire. We diversified the clinician type of our sample by asking participants for referrals toward the end of the interview. Further, we used our professional networks to contact clinicians, and we posted recruitment messages to social media, accompanied with keywords targeted toward clinicians working in cancer. A total of 59 unique recruitment emails were sent between July and December 2020. Interviews were conducted simultaneously with recruitment, as the first interview occurred in July. During this time, the number of COVID-19 cases in the state of Florida peaked in October before plateauing in December [18]. Pharmaceutical companies were also beginning to seek approval for vaccines.

Procedures

Potential participants filled out a short online form to indicate their interest and to schedule an interview. Prior to the interview, participants were provided with a statement of their rights. All interviews were completed by 1 of 3 authors (JA, CH, and CB) using a semistructured interview guide about 3 different communication topics in cancer care (secure messaging, teleoncology, and online information seeking). Questions about communication using teleoncology during the COVID-19 pandemic made up 1 of 3 sections of the interviews. Members of the research team collectively wrote the interview guide to align with our goals of understanding clinicians' perceptions of teleoncology. The clinical member of the research team (author MJM) reviewed the interview guide before it was finalized. Specific questions included asking clinicians about the challenges they encountered in moving to telehealth to communicate with patients, its advantages/disadvantages, and what strategies were developed to facilitate telehealth interactions. Interviews were conducted using the videoconferencing software Zoom and were audio-recorded and professionally transcribed.

Data Analysis

The constant comparative method [19] was utilized to analyze the interview transcripts using an inductive, thematic approach. Thematic analysis is a valuable method for examining the perspectives of different participants, highlighting similarities and differences, and generating unanticipated insights [20]. Interviews continued during data analysis until no new themes emerged and thematic saturation was achieved [21] through recurrence, repetition, and forcefulness of the data [22]. The second author (GT) uploaded all transcripts to Atlast.ti v. 22 (ATLAS.ti Scientific Software Development GmbH), a software management and analysis program. Two authors (GT and CB) conducted open coding using an adapted version of Strauss and Corbin's guidelines, [23] assigning in vivo codes. Codes were collapsed into categories, after which thematic properties were identified using axial coding. For example, each participant's interview was examined for information relevant to 1 of the

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posed inquires (ie, benefit or barrier) and Atlas.ti was used to assign a code. Codes were compared and combined to generate themes, which were then examined for text that conveyed similar messages, after which those were separated into their own group (i.e., property). Codebooks were developed for each research inquiry throughout the analytical process by the second author (GT) and were discussed with the senior author (CB) to refine themes and properties before creating finalized versions. The second author (GT) created analytical notes and memos throughout the analysis process, which increased the ability to identify poignant descriptions to illustrate themes and properties. This strategy was used to increase the trustworthiness of findings as well as promote transferability [24]. The senior author (CB) used the final codebooks to conduct closed coding of all transcripts, after which the second author (GT) validated the analysis. At this point, we shared the analysis with our clinician coauthor (MM) and study principal investigator (JA) for further validation of the results.

Results

Participant Characteristics

A total of 21 clinicians participated in the study (36% enrollment rate). Interviews averaged 44 minutes in length and resulted in 285 transcribed pages. Of the 21 participants, 13 (62%) were female; the average number of years postresidency, fellowship, or schooling among 18 (86%) participants was 8 years (range 1-33); and 3 (14%) participants were still in residency. One (5%) participant was a physician's assistant, and another (5%) was an advanced practice registered nurse. Most clinicians (n=17, 81%) were affiliated with the UFHCC, while the other 4 (19%) were employed at cancer centers in the south, northeast, and western U.S. Most clinicians primarily worked in outpatient settings, and 14 (67%) were in medical oncology departments and 7 (33%) in radiation oncology. Each participant reported that they used teleoncology with patients during the pandemic.

Our qualitative analysis revealed a total of 6 themes: 3 (50%) themes related to the benefits of teleoncology and 3 (50%) themes about the challenges of teleoncology. We describe each theme next and include thematic properties, when present, to provide a richer description of the themes. Additional exemplar quotes associated with each theme and property are in Tables 1 and 2.

Table 1. Benefits of technology.

Properties (if applicable)	Exemplar quote
Theme: teleoncology is convenient for patients	
Reduces in-person visits and travel	*A lot of our patients do travel very far to see us, and I think telemedicine can be used very effectively for visits where we don't necessarily need to see the patient in person or it's our first encounter and we want them to get more studies done before seeing us.*
Reduces financial burden	*It cuts down on the financial burden for them and having to come in the office purely to have a discussion and then for us to tell them, "You need more imaging," and then them having to come back . . . same for follow-up appointments.*
Theme: teleoncology reduces the risk of COVID-19 exposure	
__[a]	*I think the biggest advantage is being able to keep people who are at higher risk for complications from COVID at home and out of the general public.*
Theme: teleoncology helps clinicians to better "see" patients and family	
Makes patients and their environments visible	*I get to see inside their home. So, if I can tell there's a dog in the room, I usually ask them to show me their pet. I had a patient this week walk me by phone outside into her garden to see an orchid that she had blooming . . . It's a neat way to connect with them that we can't do in the clinic.*
Facilitates family member participation	*[Maybe] they can have family members present who may not otherwise be able to be present.*

[a]Not applicable.

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Table 2. Challenges of technology.

Properties (if applicable)	Exemplar quote
Theme: technical challenges affect the quality and effectiveness of teleoncology	
Internet connectivity issues	*A lot of my patients, because they're rural, don't have Wi-Fi strong enough for me to do an actual Zoom visit. It's very frustrating, because it drops so much, and it freezes. So, they've just given up, and they just come to clinic.*
Patients' unfamiliarity with telehealth technology	*The biggest challenge was literacy about technology. Most of our patients, sometimes you will get into the Zoom, they are not there, and they are waiting on you, [and] then they will call the clinic. I've been waiting on my doctor because they don't know how to navigate it.*
Theme: inability to conduct a physical exam	
__[a]	*The challenges are definitely not being able to do a physical exam because the patient is not there with you in person. You're seeing them in their environment, sitting in a chair, but you're not seeing them walk into the office. You can gather a lot by watching someone walk in and if they're struggling to walk in, those types of things.*
Theme: challenge to meet expectations about appointment times	
—	*I've noticed with Zoom, there's this expectation that I be exactly on time. And our clinic schedules face-to-face and Zoom all mixed in. So, by definition, I never see a clinic patient at the time of their appointment, because they're getting vitals. So now my 9:00 AM, I don't see till 9:20, but my 9:30 expects me to be on Zoom right at 9:30, and I just can't actually do it.*

[a]Not applicable.

Benefits of Technology

Theme 1: Teleoncology is Convenient for Patients

Teleoncology was described by clinicians to be better for the patient than in person-visits in many ways as it removed traditional demands (eg, driving to an appointment, planning) and requirements (eg, sitting in the waiting room, around others).

Reduces In-person Visits and Travel

Clinicians shared that teleoncology provided patients with an opportunity to avoid physically coming to the office/clinic to meet with providers when it was not necessary. A radiation oncologist noted that telemedicine could be an effective substitute for situations such as a first visit when more tests are warranted, general consultations, or follow-up appointments. One clinician said:

> *[Patients] who are on routine follow-ups . . . and some that are in remission who just come in every 3 or 6 months or even annually for lab work who don't have any new physical issues, physical symptoms, any new concerns, who are doing great, they really can just go get lab work done outside . . . They don't have to come into clinic.* [Participant 28]

One benefit of reducing in-person visits is not needing to travel, especially for those who live far away from the hospital, such as those who live in other parts of the state or other countries. One clinician recalled an experience where they were able to consult with a patient living in another country using teleoncology:

> *One of the patients I saw . . . was from the U.K., and that's [teleoncology] was the only way we were going to be able to see him at that time.* [Participant 59]

Reduces Financial Burden

Clinicians also described how teleoncology reduced the financial burden of coming to appointments in person. Clinicians cited travel expenses for individuals, especially those with a limited financial budget. One clinician spoke specifically about the advantage teleoncology afforded patients with fiscal issues:

> *We see those low socioeconomic groups so common and people who don't have gas money. I mean, that's huge, and obviously people that travel several hours.* [Participant 8]

Another clinician echoed this by addressing the distance some patients are required to drive to a clinic for a short appointment, saying:

> *For patients who don't have a lot of money or have transportation issues, it really saves them a visit. So, if there are things that it's just a conversation, and it really doesn't require them to drive 150 miles to have a 20-minute conversation with me, I think that's a beautiful use of telemedicine.* [Participant 2]

Theme 2: Teleoncology Reduces the Risk of COVID-19 Exposure

Teleoncology made it convenient for immunocompromised patients to avoid exposure to high-risk health care areas where COVID-19 might be present and being around the public when traveling to and from appointments. One clinician described how this form of communication enabled at-risk patients (both with cancer and in remission) to stay at home, while also highlighting that they were, and most likely would be, immunocompromised to some degree.

> *They don't have to put themselves at risk by coming into clinic, because some of these patients, they're*

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> *cancer survivors or they're cancer patients in remission and they're still at risk in terms of their immune system. To some level, they're always immunocompromised because of their treatment, so there's no reason to bring them into [the] clinic . . . So they can stay in the safety of their own home and do a quick telemedicine visit, and it's simple and they prefer that. They don't have to leave their house.* [Participant 28]

The level of concern patients had regarding exposure to external environments during the pandemic was also cited by participants. One clinician described patients' concerns and mentioned the safety this form of communication afforded immunocompromised individuals:

> *It allowed the opportunity for patients to stay home, be safe. A lot of these patients obviously are immunocompromised, and if it's just like a lab check, we can do that over Zoom. We don't need to do, like, a physical exam at that point. I think it just gives the patients peace of mind. I mean, a lot of them were very nervous, understandably, to come in. So, we're able to provide that service.* [Participant 44]

Theme 3: Teleoncology Helps Clinicians to Better "See" Patients and Family

Clinicians reported the benefit how interacting with a patient via videoconference provided them a unique opportunity to see the patient's environment and speak with caregivers or family members who could attend the online appointment.

Makes Patients and Their Environments Visible

Clinicians described the importance of "seeing" patients, as opposed to only talking to them over the phone. One participant compared it to doing a home visit in that it allowed them to assess whether the patient was physically well. Another oncologist recalled how a virtual visit with a patient helped their decision making:

> *I had a patient who did a telehealth visit with me from her bed, because she couldn't get out of bed. And she wouldn't tell me that. But the fact that she did this visit with me, laid up in bed, and hadn't gotten ready, it told me so much about what was going on with her healthwise that it was sort of invaluable information for me to make decisions. And then just seeing where they live and what their living situation is like, and you can just get so much information from a telehealth visit that you'll never get from an in-person visit in the clinic.* [Participant 2]

Clinicians also said that viewing patients' living conditions provided an opportunity to make connections and form rapport that they might not have been able to do in a traditional setting. One clinician spoke about seeing pictures and other items inside of a house and striking up conversations with the patient. They said:

> *It was nice to have conversations about pictures that they had in their house, or items that they had in their house that I found interesting, and it was always a nice way to get to know people on a personal level, and kind of develop a rapport with them.* [Participant 13]

Another recalled having a virtual visit with a patient who was outside, and noticed animals in the background, allowing them to form a connection with the patient. They said:

> *One of my patients did it from outside, because that was the only place he had a cell signal, and so you could see all his chickens and his pig in the background. And I have chickens, too. So, I did talk about the chickens, and I got to meet his pig, and that was just a really lovely connection that I wouldn't have really had with the patient.* [Participant 6]

Facilitates Family Member Participation

Clinicians noted that teleoncology provided caregivers and family members who might not be able to attend in-person visits an opportunity to engage in discussions. One clinician illustrated this by saying:

> *It gave us a chance to get a sneak peek into a patient's home, which we never necessarily saw before, so patients who didn't have caregivers ever accompany them sometimes they were in the chair next to them at the table.* [Participant 7]

Challenges of Technology

Theme 1: Technical Challenges Affect the Quality and Effectiveness of Teleoncology

Clinicians described how a variety of technical challenges hindered the ability to conduct a clinical appointment over Zoom. This included internet issues and low confidence using computer applications.

Internet Connectivity Issues

Clinicians described instances when a virtual visit would be interrupted due to low bandwidth or an unstable connection. Clinicians frequently expressed how low bandwidth contributed to unstable connections for patients who lived in rural areas, which resulted in dropped calls, freezing screens, and delays.

> *The patients that I was doing telemedicine with live in kind of rural, outlying areas, and so I found that we could get connected . . . it took a little bit of time, and then there were lots of delays. And in a couple of situations, people got cut off, and we had to log back in.* [Participant 2]

Patients' Unfamiliarity With Telehealth Technology

Clinicians described how patients' lack of familiarity using technology (eg, Zoom, installing applications) negatively impacted communication. As 1 clinician described:

> *Everybody wasn't able to use Zoom as effectively initially, and so you'd have situations where people couldn't log in, they couldn't be heard or seen because the program wasn't working correctly, and so it was just kind of frustrating some people, so they may not necessarily show up for an appointment because they*

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> *don't want to have to deal with Zoom.* [Participant 58]

Clinicians cited patients' age as a contributing factor to the lack of familiarity with technology. They noted that elderly patients were not always "technologically savvy" with telemedicine services, such as Zoom or online portals, as illustrated by the following recollections:

> *The biggest challenge is that we have, generally speaking, an elderly population of patients, some of [whom] are very tech savvy and can Facetime or Zoom or use email. But there was some disparity that was created because some patients were not used to using technology in that way.* [Participant 59]

> *I see a particular group of patients [who] are typically elderly, and might not be technologically savvy, in order to know how to access the telehealth portal. And that became a little bit challenging, and it would have to require the help of either me or my staff to get them connected.* [Participant 13]

Theme 2: Inability to Conduct a Physical Exam

Not physically being together meant that physical exams were unable to be performed. One clinician explained how not being able to conduct a physical exam was a particular challenge with cancer patients:

> *You have to see these patients and be able to assess their fitness for chemotherapy, and that takes the ability to actually lay eyes on them and examine them and really teach them. A lot of the things we ask of our patients are not easy requests, and it's also part of the care is also emotional support. And sometimes, that doesn't translate as well online. So in order to give the comprehensive care that they need, then visits are important.* [Participant 9]

Another clinician remarked about the significance of being with a patient face-to-face. Using teleoncology, the clinician acknowledged that they were unable to see the patient walk into the office. Information gathering can occur by observing if a patient is struggling to walk or by the way they position themselves on the examination table.

Theme 3: Challenge to Meet Expectations About Appointment Times

Without in-person visits, clinicians also described that patients' expectations and behaviors had changed since using teleoncology services. For example, 1 oncologist said that patients expected them to be exactly on time:

> *Our clinic schedules [include] face-to-face and Zoom all mixed in. So, by definition, I never see a clinic patient at the time of their appointment, because they're getting vitals. So now my 9:00 A.M., I don't see until 9:20, but my 9:30 expects me to be on Zoom right at 9:30, and I just can't actually do it.* [Participant 6]

Other oncologists noted that when they were not on time, some patients left the videoconference. One oncologist shared how their expectations of patients waiting on Zoom were much different from the reality, saying:

> *I thought when we started using it like, "This'll be great when people have to wait. If I'm running late, wouldn't you rather wait in your own home, and you'll just Zoom on?" And not so much. There's not a great way to let people know how long they're going to be waiting. That system is not really well worked out, and so I think that's kind of annoying for patients. Well, and for me too. They'll Zoom on. If you're late, they're gonna Zoom off. You got to get them back. That's kind of cumbersome.* [Participant 8]

Discussion

Principal Findings

The COVID-19 pandemic led to a major shift in the way cancer care was provided to patients for a sustained period. As there have been calls for teleoncology to be more present in cancer care [5,6], it is important to understand this almost universal experience of teleoncology from the perspective of clinicians delivering care. After conducting 21 interviews with oncology clinicians about their experiences pivoting to teleoncology during COVID-19, we found that utilization of the technology has many benefits but also has several challenges to be overcome if it is to continue as a viable option for appointments and consultations. Clinicians believed that teleoncology has nonmedical benefits for patients, such as reducing travel time and expenses related to the consultation, as well as medical benefits, such as limiting COVID-19 exposure and allowing clinicians to get a better sense of the patients' lifestyle, environment, and incorporating family members. Challenges also comprised nonmedical and medical issues. Nonmedical factors were technology related, such as problems with internet connectivity and lack of familiarity with videoconferencing technology. However, clinicians perceived shortcomings in teleoncology because they could not have physical contact with patients, which inhibited their ability to conduct a physical exam. Further, instances occurred in which patients were disappointed and frustrated that clinicians were late to the Zoom appointment.

This study adds to the growing literature on teleoncology by highlighting the perspective of oncology clinicians. The previous literature about teleoncology has focused on the experience of using the technology as a tool to reach patients in rural settings and developing countries [25-28]. Our findings align with the literature emphasizing the benefits of teleoncology to reduce travel time and costs, but in the case of COVID-19, teleoncology was mandated as the primary method of care for patients with cancer. Adoption of new technology can be slow, especially in health organizations because organizational (eg cost, complexity, impact) and individual factors (eg age, attitude) determine when and if innovations are accepted [29]. Due to the pandemic, health systems decided to universally adopt teleoncology, even though there was uncertainty among end users (ie, clinicians), otherwise known as forced adoption [30]. As a result, clinicians in our study dealt with the benefits and challenges of teleoncology concurrently, without the ability to address and fix challenges.

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However, being compelled to use teleoncology pointed out a benefit that seems to be missing in the previous literature, that of the ability to "see" the patient and their family. In this case, "seeing" could mean several things: (1) viewing the patient's health and symptoms (as opposed to telephone only); (2) observing the patient in their home environment, which further allowed for better rapport building and connection; and (3) witnessing the patient within the context of their family situation, as family members who could not normally attend were able to. Interestingly, 1 clinician discussed how teleoncology was akin to a home visit because it was an opportunity to observe the patient in their own environment. Knowledge of a patient's physical living space could benefit clinicians in providing care [31]. In all cases, the ability to "see" had the potential to improve care for the patient by better understanding their situation.

Challenges of teleoncology were noted as including technical difficulties, which are well established in the literature. A recent study among clinicians found that poor internet connectivity is the biggest barrier to telemedicine [32]. Lack of access to technology, which enables the use of teleoncology, is also a significant issue that has implications for health equity in cancer care delivery. Compared with younger patients, older patients with cancer are less likely to have an email address or own a smartphone and are less likely to use a patient portal to communicate with their oncology care team [33]. In addition, patients faced similar hurdles as clinicians to forced adoption of teleoncology. Digital literacy—the awareness, attitude, and ability to appropriately use digital tools and facilities to identify, access, manage, and construct new knowledge and communicate with others [34]—is a major factor that has widened the digital divide. Older adults (65+ years old) have the lowest adoption rates for using new technologies [35]. However, internet adoption among older adults has risen steadily over the past decade and a half [36]. An intervention that trained older adults to use technological devices found improvement in technology confidence and a significant increase in technology use [37]. Other than an email with instructions, patients received little guidance about shifting to teleoncology.

Another challenge faced by clinicians was the inability to conduct physical examinations. Although tools such as a weight scale, blood pressure cuff, pulse oximeter, and thermometer can be administered by patients while using telemedicine, such tools are sometimes not covered by insurance and may be prone to errors due to lack of calibration and patients' inexperience [38]. It is important for clinicians to physically examine patients, but it is not necessary for certain types of appointments. As clinicians in our study acknowledged, teleoncology is beneficial for follow-ups and instances when patients are not experiencing any discomfort.

Interestingly, 1 of the themes that emerged was a different expectation from patients about how appointment start times should be managed. Clinicians observed that patients assumed the clinician would be present at the start of the Zoom appointment, even though it is commonplace for patients to wait for the clinician during in-person appointments. Patient satisfaction is negatively impacted by longer wait times and affects perceptions of information, instructions, and the overall treatment provided by clinicians [39]. Among patients with cancer, over 80% in an outpatient oncology clinic felt that waiting for their appointment had an emotional cost [40]. Further, over one-quarter of patients suffered a major emotional impact by seeing other sick people in the waiting room [40]. Although better coordination and communication is necessary when scheduling teleoncology appointments, patients do have the benefit of waiting in their home rather than in the clinic. If patients were made aware of possible delays or received periodic updates about the status of their appointment, perhaps fewer patients would abandon the Zoom appointment. There is the potential to damage the clinician-patient relationship when clinicians are delayed. Uncertainty and lack of communication between the patient and the health care team can have negative implications, but keeping patients informed and expressing empathy are ways of improving the interaction [41,42].

Implications of the Study

There are several practical implications from this study for those working in clinical settings as either clinicians or administrators. First, clinicians should receive training about communicating effectively with patients using teleoncology. Our study identified challenges to using teleoncology that could be remedied with slight modifications to clinicians' behavior. For instance, patients satisfied with encounters using telemedicine appreciated relational experiences with clinicians and when an effort was made toward building a patient-centered relationship [43]. Clinicians should also look at the camera to ensure good eye contact and foster rapport and trust [44]. Training can also include how to involve family members present on-screen and methods to managing appointment times. Second, the health care team can inform patients when scheduling about what to expect before the appointment begins. Notifying patients of potential delays and having clinicians update patients during appointments while they are waiting can reduce uncertainty. While patients are waiting, health care teams can use the opportunity to emphasize the importance of health promotion through COVID-19 risk reduction by playing videos and other educational content. Lastly, it is important to ensure that patients are prepared for the appointment by testing out the technology in advance and having flexibility about what type of technology they can use. Since the Health Insurance Portability and Accountability Act (HIPAA) relaxed its guidelines for COVID-19, tools such as Apple FaceTime, Facebook Messenger video chat, Google Hangouts video, and Skype can be utilized [44]. Patients should be offered the choice of technology to use for teleoncology in order to avoid downloading and learning new applications. For teleoncology to be successful and a valid method of care delivery, ultimately, the responsibility falls on the health care system to better accommodate the technology than placing the burden on clinicians. However, the rapid increase in teleoncology visits during the pandemic has revealed that it should have a larger role postpandemic. In 2021, at least 30 states considered legislation to revise telehealth coverage standards [45]. In addition to ensuring that all patients can access teleoncology services, including telehealth as part of routine follow-up care has been recommended because it allows for efficient discussions of laboratory and imaging results, as well as side effect management [16].

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Limitations

Although we attempted to diversify our sample by recruiting clinicians from different health systems, the majority of participants were from 1 health system. Therefore, our results may not extend beyond the health system and be generalized in other contexts. There may also be the possibility of selection bias, as participants in our study volunteered. Most participants were oncologists, but understanding the experiences of other types of oncology clinicians is critical. Interviews took place toward the end of 2020 after teleoncology use spiked in the previous months. At the time of the interviews, teleoncology was relied upon less frequently. Developments related to COVID-19 have caused frequent shifts in health care protocols, which highlights the need for further research to examine the long-term implications of teleoncology.

Conclusion

We interviewed 21 cancer clinicians during the COVID-19 pandemic to understand the benefits and challenges of using teleoncology to replace in-person appointments. The rapid adoption of teleoncology resulted in several obstacles, such as issues around internet connectivity and miscommunication about appointment times. Benefits included reduced travel time for patients and limiting their exposure to COVID-19. Clinicians appreciated the ability to learn more about patients by observing their living conditions, which provided insights into the patient's lifestyle. Future work is warranted to explore the attitudes and perceptions of patients, along with clinicians, in various types of cancers to understand how the technology is adapted to different types of diseases. Future research should also include family members and caregivers to understand their role in the facilitation of teleoncology and how their involvement can alter depending on the type of visit.

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Conflicts of Interest

None declared.

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Abbreviations

UFHCC: University of Florida Health Cancer Center

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