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Contents

Review

Examining Patient Engagement in Chatbot Development Approaches for Healthy Lifestyle and Mental Wellness Interventions: Scoping Review

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Abstract

Background: Chatbots are growing in popularity as they offer a range of potential benefits to end users and service providers.

Objective: Our scoping review aimed to explore studies that used 2-way chatbots to support healthy eating, physical activity, and mental wellness interventions. Our objectives were to report the nontechnical (eg, unrelated to software development) approaches for chatbot development and to examine the level of patient engagement in these reported approaches.

Methods: Our team conducted a scoping review following the framework proposed by Arksey and O'Malley. Nine electronic databases were searched in July 2022. Studies were selected based on our inclusion and exclusion criteria. Data were then extracted and patient involvement was assessed.

Results: 16 studies were included in this review. We report several approaches to chatbot development, assess patient involvement where possible, and reveal the limited detail available on reporting of patient involvement in the chatbot implementation process. The reported approaches for development included: collaboration with knowledge experts, co-design workshops, patient interviews, prototype testing, the Wizard of Oz (WoZ) procedure, and literature review. Reporting of patient involvement in development was limited; only 3 of the 16 included studies contained sufficient information to evaluate patient engagement using the Guidance for Reporting Involvement of Patients and Public (GRIPP2).

Conclusions: The approaches reported in this review and the identified limitations can guide the inclusion of patient engagement and the improved documentation of engagement in the chatbot development process for future health care research. Given the importance of end user involvement in chatbot development, we hope that future research will more systematically report on chatbot development and more consistently and actively engage patients in the codevelopment process.

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KEYWORDS

chatbots; virtual assistants; patient involvement; patient engagement; codevelopment

Introduction

Growing evidence supports the use of digital technology in healthy eating, physical activity, and mental wellness interventions. Several systematic reviews on these digital health interventions (DHIs) have identified their promise in managing chronic diseases [1-6]. Specifically, DHIs have proven impacts on reducing risk factors for chronic diseases [3,4] by increasing

physical activity, reducing body mass index [6], and improving patient psychosocial well-being [2]. Further, DHIs can help overcome barriers to access to mental health support for individuals with chronic conditions [1]. Although these DHIs are useful in vulnerable chronic disease populations [5,7], they face challenges, including limited user adoption, low engagement, and high attrition rates [8-11].

Chatbots are artificial intelligence (AI) programs that converse with humans through natural language in text or speech [12]. There is a growing body of evidence that the integration of chatbots into DHIs may provide support [13-17] by increasing patient engagement [13], intervention adherence [13], and the acceptability and efficacy of lifestyle and wellness interventions [15-17]. Additionally, chatbots offer a range of potential benefits to end users and service providers, most notably allowing for more scalable, cost-efficient, and interactive solutions [12].

Although developments in AI and computer science have improved the ability of chatbots to mimic human agents, the acquisition of a relevant data set with which to train chatbots remains challenging. User-centered design with public and patient involvement (PPI) may offer a potential solution [18-20]. By engaging key stakeholders, PPI can help produce better-quality interventions relevant to end users's needs [18], resulting in benefits such as increasing intervention acceptability, effectiveness, and sustainability [19]. Drawing on evidence across other digital health care innovations, the proposed benefits of PPI fundamentally include the development of interventions that are both usable by and relevant to patients [19]. Recognizing the limited data available to guide the role of PPI in digital health innovation, experts have called for the meaningful involvement of patients from the beginning of the development process to allow for the cocreation of relevant, valuable, and acceptable digital health solutions [20].

This scoping review aimed to map the literature on studies using chatbots to engage in 2-way natural language interaction (voice- or text-based input) to aid the delivery of healthy eating, physical activity, and mental wellness interventions. The specific objectives of this review were: (1) to report the nontechnical (eg, unrelated to software development) approaches for chatbot creation and (2) to examine the level of patient engagement in these reported approaches. Although the technical software development steps are essential to creating chatbots, this review focused on the nontechnical approaches for chatbot development as these are less explored and more likely to involve patient participation. To our knowledge, this is the first scoping review to systematically explore these objectives.

Methods

Study Design

This scoping review was conducted using the framework proposed by Arksey and O'Malley [21] and later refined by Levac et al [22]. The Arksey and O'Malley framework consists of the following five steps: (1) identify a research question, (2) identify relevant studies, (3) select studies, (4) chart the data, and (5) summarize and report the results [21]. Two research questions guided the review:

1. Outside of the technical software development processes, what approaches are described for the development of chatbots that support healthy eating, physical activity, and mental wellness interventions?
2. What is the extent of patient engagement in these approaches?

Study Team

Our multidisciplinary study team included 2 graduate student researchers (CS and CC), a health sciences librarian (SC), 2 postdoctoral fellows with backgrounds in clinical care and scoping reviews (ND and AH), a professor of medicine (PT), a professor of physiotherapy (MM), and a professor of computing science (ES).

Search Strategy

A health sciences research librarian (SC) was consulted to develop a search strategy that used concepts from our research questions. The search strategy (Textbox 1) included a combination of subject headings and keywords, including health, chatbots, and lifestyle or wellness components. Searches were adjusted appropriately for each database. Nine electronic databases were searched in July 2022 including OVID MEDLINE, OVID Embase, OVID PsycINFO, EBSCO CINAHL, Scopus, IEEE Explore, Proquest Dissertations and Theses Full Text, Cochrane Library, and PROSPERO (International Prospective Register of Systematic Reviews). No publication date limit was applied to the search, as the literature on chatbots and virtual conversation agents is naturally self-limiting. After conducting the search, the results were imported into Covidence systematic review management software and duplicates were removed [23]. Covidence is a "web-based collaboration software platform that streamlines the production of systematic and other literature reviews" [23]. The full text of the search strategy is in Multimedia Appendix 1.

Textbox 1. Search strategy used for OVID PsycINFO database.

Searches

1. (chatbot* or “im bot” or “im bots” or “instant message bot*” or “conversational agent*” or “virtual agent*”).mp.
2. *“Diets”/
3. *“Health Promotion”/
4. *“Intervention”/
5. *“Physical Activity”/
6. “Nutrition”!
7. “Weight Loss”!
8. “Sedentary Behavior”/
9. (lifestyle* or health* or medic* or nursing or nurse* or disabilit* or elder* or “senior citizen*” or patient* or exercise or “physical activit*” or motivational).mp.
10. 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9
11. 1 and 10

Eligibility Criteria

Included publications were those written in English and published in peer-reviewed journals. Included studies all had an intervention supporting healthy eating, physical activity, and mental well-being. All studies required a chatbot that communicated with users through a 2-way natural language interaction. Inclusion criteria for participants consisted of adolescents (age >10 years old) as defined by the World Health Organization [24] or adult populations. Studies were excluded if they involved additional technologies or chatbot service delivery beyond the scope of this review (ie, embodied conversation agents, humanoid and social robots, wearable technology, Internet of Things (IoT), virtual avatars, interactive voice assistants, or chatbots delivering therapy to clients). Studies were also excluded if they only described an intervention but did not conduct or test one. Chatbots designed to replace a therapist’s role were excluded, as were papers that did not present original results (ie, reviews and protocol papers). Randomized controlled trials (RCTs) were included in recognition that they often contain valuable insights into the development process, particularly when the authors did not publish a formative manuscript.

Study Selection

Titles and abstracts of the retrieved articles were reviewed independently by 2 researchers (CS and CC) based on the inclusion and exclusion criteria described above. Both reviewers met throughout the title and abstract screening stage to discuss and resolve conflicts through consensus. A third reviewer (ND or AH) was consulted for consensus. The remaining articles advanced to the full-text screening stage. The excluded articles were tagged with reasons for exclusion derived from our exclusion criteria. After independent full-text screening, both reviewers met to resolve any inclusion or exclusion and exclusion reason conflicts. Interrater reliability was assessed using the Cohen κ [25]. For the included articles, an additional literature search was carried out using the involved authors, chatbot details, and reference lists to determine whether the previous formative papers that described the chatbot development had been published.

Data Extraction

One reviewer (CS) extracted the data from included articles using a standardized Microsoft Excel form. General and specific data were extracted, including author, publication year, journal, study setting, study design, sample size, participant demographics (age, sex, and chronic disease where applicable), intervention type, chatbot type, chatbot development approaches, and assessment of patient involvement in development.

Patient involvement was assessed using the Guidance for Reporting Involvement of Patients and Public (GRIPP2) short-form checklist [26]. The GRIPP2 checklist was applicable for our objectives as it was designed to enhance the quality of patient and public involvement (PPI) reporting in health technology assessment and health research [26], and because it could be used retrospectively to measure the quality of PPI reporting in publications and reports [27]. Table 1 depicts the GRIPP2 checklist as we used it to assess PPI in chatbot development. The GRIPP2 awards points across 5 items that describe public engagement and involvement.

Table 1. How the Guidance for Reporting Involvement of Patients and Public (GRIPP2) reporting checklist was used to grade patient and public involvement in chatbot nontechnical development.[a]

Section and topic	Specifics for engagement in chatbot-related development
1. Aim	Report the aim of PPI[b] in chatbot development
2. Methods	Provide a clear description of the methods used for PPI in chatbot development
3. Study results	Outcomes: Report the results of PPI in chatbot development, including both positive and negative outcomes
4. Discussion and conclusions	Outcomes: Comment on the extent to which PPI influenced chatbot development overall. Describe positive and negative effects
5. Reflections or critical perspective	Comment critically on chatbot development, reflecting on the things that went well and those that did not, so others can learn from this experience

[a]Adapted from Staniszewska et al [27].

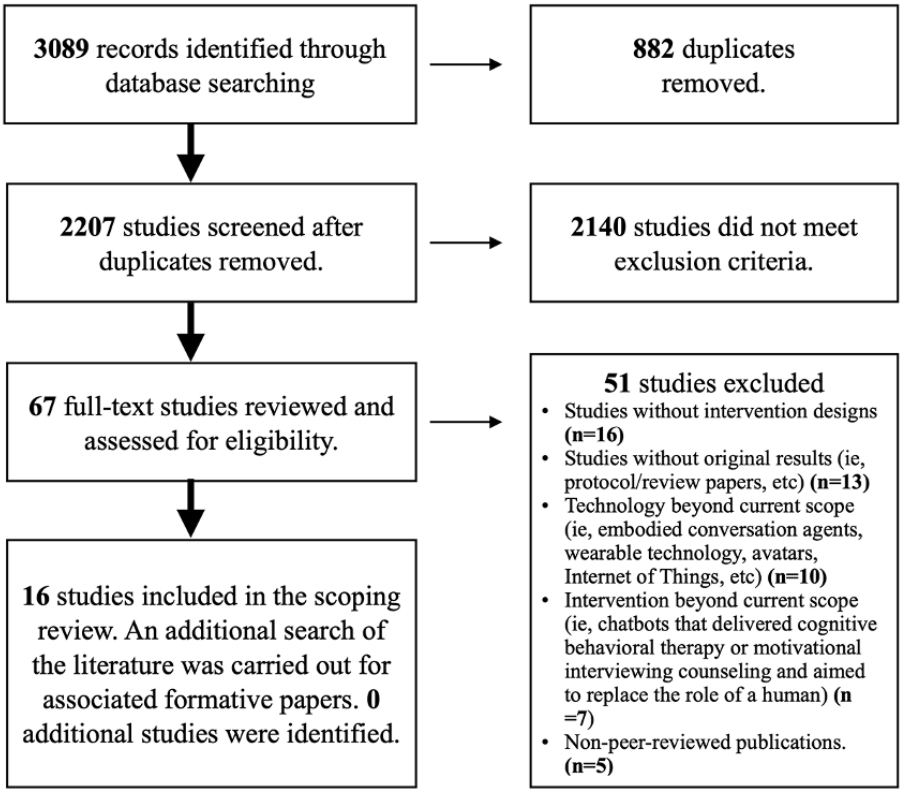
[b]PPI: patient and public involvement.

Results

Search Results

Figure 1 shows the search results; 3089 publications were retrieved from the database searches, and 882 duplicates were removed, leaving 2207 studies to screen. At the title and abstract screening stage, there was "fair" agreement between reviewers (Cohen κ=0.309, proportionate agreement=0.967). After completing the title and abstract screening, 2140 publications were removed as they did not meet the inclusion criteria. Reading the full text of the remaining 67 publications resulted in a further 51 publications being excluded, with the exclusion reasons documented in Figure 1. At the full-text review stage, there was "almost perfect" agreement (Cohen κ=0.843, proportionate agreement=0.941). In total, 16 publications were included in this review.

Figure 1. PRISMA (Preferred Reporting Items for Systematic reviews and Meta-Analyses) flow diagram of included and excluded studies.



Description of Included Studies

Table 2 shows the description of the included studies and their chatbot interventions. The included studies were conducted in 4 countries, with 50% (8/16) of the studies conducted in Canada [28-35]. Six studies were conducted in Switzerland [36-41], 1 study was conducted in Saudi Arabia [42], and 1 study was conducted in Korea [43]. The majority of the studies (14/16) were conducted in a health care setting [28-40,43], with the remaining 2 studies in a computing science setting [41,42]. All but one of the included studies [31] were published in 2020 or later.

Table 2. Descriptive summary of included studies, chatbots, and their development.

Study and country	Study type	Chatbot intervention	Approaches for development	Identified development approaches	Patient engagement (GRIPP2[a])
Alghamdi et al [42], Saudi Arabia	Randomized controlled trial	Text-based nutrition chatbot for patients with celiac disease	• Literature review of existing health behavior change models. Investigated the pros and cons of each model to guide development of a health behavior change model to structure the chatbot's content • Interviews with expert users (from patient population diagnosed with celiac disease 4+ years ago, patient's parent, dietitian supervising patient for 4+ years, gastroenterologist treating celiac disease patient for 4+ years) • Questionnaires for patients with celiac disease to understand symptoms and technology use preferences	• Literature review • Patient interviews • Collaboration with knowledge experts	Unable to assess
Davis et al [36], Switzerland	Nonrandomized experimental study	Text-based exercise and nutrition chatbot	• Development outsourced to a software company; did not report any steps taken for development	• None identified	Unable to assess
Dhinagaran et al [28], Canada	Feasibility study	Text-based exercise, nutrition, and wellness chatbot for patients with diabetes	• Needs assessment conducted in an earlier publication • Literature review of systematic reviews and clinical guidelines for evidence-based content development to develop content • After a 4-week pilot feasibility study, conducted follow-up interviews to understand patient views of the chatbot and to gain ideas for improvement	• Literature review • Patient interviews	Unable to assess
Figueroa et al [37], Switzerland	User design study	Text-based exercise chatbot	• Qualitative interviews during prototype testing to assess opinions and knowledge of chatbots as personal health coaches, technology use, digital literacy, and privacy considerations of chatbots in general • Wizard of Oz procedure. Participants completed a 20-minute SMS text messaging conversation with a simulated chatbot • Chatbot prototype testing. Participants texted the prototype for 10-20 minutes. Directly after the testing period, participants had a semistructured interview via videoconference regarding the chatbot's ease of use, usefulness, humanness, and sustainability • Co-design workshop for participants to take part in development of ideas for chatbot use and design. These workshops were held over Zoom and ideas were visualized on Google Jamboard	• Patient interviews • Wizard of Oz procedure • Prototype testing • Co-design workshops	Met criteria on GRIPP2 checklist points 2, 4, and 5. Provided a clear description of the methods used for PPI[b], commented on how PPI influenced the study, and on successful and unsuccessful aspects of the study relating to PPI
Gabrielli et al [29], Canada	Proof-of-concept study, mixed methods	Text-based wellness chatbot	• Intervention design. The intervention, targets, and components were defined to specify clinically relevant effects on users and to refine the intervention components. This was done by a team of 3 clinical psychologists, 2 users, and behavior change experts • Preliminary testing. A proof-of-concept implementation of the digital intervention and chatbot to examine engagement and effectiveness with a convenience sample of university students	• Collaboration with knowledge experts • Prototype testing	Unable to assess

Study and country	Study type	Chatbot intervention	Approaches for development	Identified development approaches	Patient engagement (GRIPP2[a])
Gabrielli et al [30], Canada	Pilot, co-design study	Text-based wellness chatbot	• Co-design workshop. The students used and commented on a prototyped session of the chatbot intervention to collect their needs and preferences on the following: the chatbot's look and feel, the type of content and duration of the session, their unmet expectations regarding the prototype, and suggested improvements • Feasibility test. This formative study aimed to assess the perceived value of the coaching intervention and to check the user experience with intervention to refine content	• Co-design workshops	Met criteria on GRIPP2 checklist point 2. Provided a clear description of the methods used for PPI
Greer et al [31], Canada	Randomized controlled trial	Text-based wellness chatbot for patients with cancer	• Literature review of the Stress and Coping theory and the Broaden-and-Build theory of positive emotion and focused on the teaching and practice of 8 positive psychological skills. Created lessons based on this review for the chatbot to deliver • Interviews and focus groups as formative work to refine content for the chatbot format and inform adaptation for delivery to a young user base with a shared experience of cancer treatment	• Literature review • Patient interviews	Unable to assess
Issom et al [38], Switzerland	Usability study	Text-based exercise, nutrition, and wellness chatbot for patients with SCD[c]	• Literature review of evidence-based knowledge of SCD self-management, in addition to consulting the World Health Organization's handbooks on how to implement text-based mHealth interventions to help with dialogue design	• Literature review	Unable to assess
Krishnakumar et al [32], Canada	Nonrandomized experimental study	Text-based exercise and nutrition chatbot for patients with type 2 diabetes mellitus	• Literature review to develop a lesson plan of the program. This was based on the American Association of Diabetes Educators's AADE7 self-care behaviors	• Literature review	Unable to assess
Larbi et al [39], Switzerland	Usability study	Text-based exercise chatbot	• Literature review of behavior change interventions • Summarized and briefly reported 4 steps in development: strategy planning, design, implementation, and testing. As part of strategy planning, psychology and public health experts were interviewed • Also stated that the development of the prototype involved 3 steps: requirement analysis, concept development, and implementation. Reporting did not go into any further detail	• Literature review	Unable to assess
Maenhout et al [40], Switzerland	Development pilot study	Text-based exercise, nutrition, and wellness chatbot	• Intervention planning through a scoping review of literature, conducting focus groups, and consulting web-based chat threads for a youth helpline. Focus groups addressed: content preferences, design preferences, questions that the chatbot would be asked, and answers that were expected from the chatbot • Intervention optimization through conducting a log data analysis during pretesting. A prototype of the chatbot was developed and pretested by the target users. The prototype was developed based upon guidance from phase 1 focus groups. Conversation logs were closely monitored to refine and fine-tune the chatbot. A question list was formed at the end of this prototype testing phase, 37 new (and practical) questions originated that were not covered in the chat threads and focus groups	• Literature review • Patient interviews • Prototype testing	Met criteria on GRIPP2 checklist point 2

Study and country	Study type	Chatbot intervention	Approaches for development	Identified development approaches	Patient engagement (GRIPP2[a])
Maher et al [33], Canada	Proof-of-concept study	Text-based exercise and nutrition chatbot	• Did not report how the chatbot was developed; the methods section described how the pilot study was conducted	• None identified	Unable to assess
Pecune et al [41], Switzerland	Nonrandomized experimental study	Text-based nutrition chatbot	• Literature review of persuasive systems, recommender systems, and food-related experiments • Collected a food database by regrouping the 40 ingredients that people most frequently cook and eat for dinner. These data were collected from hundreds of participants through questionnaires • Completed a pilot study to determine what the critical elements are for recipe recommendation systems. Also, completed this quasi-experimental study to understand the efficacy of different chatbot characteristics with the target end user group	• Literature review	Unable to assess
Piao et al [43], Korea	Usability study	Text-based exercise chatbot	• Needs assessment through web-based surveys to assess daily routines of office workers (the target group). This was used to determine daily activities that were measurable and easy to execute. These became a part of the goal setting in the intervention • Chatbot design was guided through a review of the literature and to determine a theoretical model for the chatbot's basis: the habit formation model • Conducted this formative usability test prior to the randomized controlled trials below to identify issues and make revisions	• Literature review • Prototype testing	Unable to assess
Piao et al [35], Canada	Randomized controlled trial	Text-based exercise chatbot	• Literature review of extrinsic and intrinsic reward systems • Steps for development were documented in the usability study described above	• Literature review	Unable to assess
To et al [34], Canada	Nonrandomized experimental study	Text-based exercise chatbot	• Development was outsourced for technical development by SmartAI. Did not report if the research team was involved in any other steps for development	• None identified	Unable to assess

[a]GRIPP2: Guidance for Reporting Involvement of Patients and Public.

[b]PPI: patient and public involvement.

[c]SCD: sickle cell disease.

Study Design and Interventions

Three of the included studies were RCTs [31,35,42], 4 were nonrandomized experimental studies [32,34,36,41], 3 were user-design and development studies [30,37,40], 3 were usability studies [38,39,43], 1 was a feasibility study [28], and 2 were proof-of-concept studies [29,33].

Fifteen of the 16 included studies reported the sample size; sample sizes ranged from 18 to 116 participants [34,37]. Participants' age ranged from 12 to 69 years, with most participants being younger than 50 years old. When a specific chronic disease group was described, populations included patients with celiac disease [42], diabetes [28,32], cancer [31], and sickle cell disease [38]. Where reported, the inclusion of female participants ranged from 31.4% to 100% [37]. Five studies involved an exercise intervention [34,35,37,39,43]. Three studies included a mental wellness intervention for healthy coping, life skill coaching, and positive psychology skill building [29-31]. Two studies evaluated a nutrition intervention [41,42]. The remaining interventions combined exercise, nutrition, and mental wellness components [28,32,33,36,38,40]. Across all reviewed articles, the chatbots communicated with users through text.

Study Findings

There were several approaches used to guide the development and training of chatbots. In 3 of the included studies, the nonsoftware development approaches for chatbot development were not documented; therefore, no approaches were identified [33,34,36]. Thirteen studies reported approaches taken for chatbot development, with most studies reporting multiple approaches [28-32,35,37-43]. In 4 of the 13 studies, patients were engaged as knowledge experts or participants in co-design workshops [29,30,37,42]. In 6 of the 13 studies, patients were involved in the study as research participants and, as part of the study outcomes, were invited to share their views through

interviews, prototype testing, and the Wizard of Oz (WoZ) procedure [28,31,37,40,42,43]. Ten of these 13 studies used a literature review, an approach that did not involve patients [28,31,32,35,38-43]. Notably, 7 of the 16 included studies were already at a more advanced stage of chatbot development, focusing on evaluating interventions and usage instead of focusing on the development process itself [31,32,34-36,41,42]. Within these studies, researchers often briefly described their overall approaches but did not go into detailed steps or explain why those steps were considered important. This did range from study to study. In 1 nonrandomized experimental study, it was reported that development was outsourced to a software company without further details regarding the process [36]. In contrast, 1 RCT effectively described the formative work their team did working with patients to refine content through interviews and focus groups [31]. However, the degree of utilization and success of the development strategy was not discussed [31]. Although we searched the literature for formative papers that preceded the included papers, no additional studies were identified using this approach (Figure 1). These nontechnical development approaches are listed and described in more detail below.

Collaboration With Patient and Clinician Partners as Knowledge Experts

During the early stages of chatbot planning, 2 studies consulted experts for chatbot development [29,42]. In both studies, patient partners were recognized as knowledge experts and included as part of the research team [29,42]. In the study with a nutrition chatbot for a celiac disease patient group, patients were recognized as experts alongside health care professionals, including dietitians and gastroenterologists [42]. In the mental wellness study, a team of 3 clinical psychologists took part in chatbot intervention development and content refinement alongside 2 users and a group of behavior change experts; this iterative process was used to adapt the chatbot's intervention program, and audiovisual content to user needs through a clinical lens [29].

Co-design Workshops

Two studies used co-design workshops to allow patients to creatively engage in the development of content ideas, chatbot design, chatbot style elements, and chatbot use [30,37]. One study invited participants to collaborate and develop ideas together with the research team over Zoom (a web-based communication platform; Zoom Video Communications, Inc) by visualizing ideas on Google Jamboard software (a web-based whiteboard for idea sharing) [37]. Another study invited patients to use a prototyped session with the chatbot to collect their needs, content preferences, stylistic ideas, and suggestions for improvements [30].

Interviews With Patients

In 5 studies, patient interviews were conducted beforehand to guide chatbot development by exploring patient needs, perceptions, and experiences with chatbot use and healthy living [28,31,37,40,42]. In 1 study, interviews were administered during prototype testing and analyzed qualitatively [37]. Another study conducted this formative work through focus groups and interviews to collect information from young adults treated for cancer, the target end user population [31]. This information was then used to guide chatbot content development within a patient-centered lens. Follow-up interviews were conducted after interventions or chatbot exposure [28,40]. Questionnaires and surveys were also used in addition to interviews to collect similar information from patients [28,42].

Prototype Testing

Many included studies were nonexperimental or pilot studies used to assess the feasibility and measure usability. These formative studies can be considered a step for development before releasing and testing a mature chatbot in an RCT. For example, 1 study using a chatbot for an exercise intervention organized a 3-week formative usability study [43] to identify issues and make revisions before conducting an RCT [35].

WoZ Procedure

One study used the WoZ procedure [37] (where the technology is controlled by a human interface in chatbot development) as a step in their chatbot development. This procedure is administered by engaging participants in a 20-minute conversation with a simulated chatbot that was not automated but controlled manually by a researcher answering questions on the back end [37]. This step was developed to understand how the chatbot should interact with humans in a natural setting and to collect content-related information directly from participants [37].

Use of Existing Literature to Gain Evidence-Based Knowledge for Development

In 10 studies, initial literature reviews were completed to gain evidence-based knowledge to guide chatbot development [28,31,32,35,38-43]. In 3 of these 10 studies, a literature review was used to develop content from evidence-based sources, including self-management practices, clinical guidelines, and systematic reviews [28,32,38]. A mental wellness study incorporated this step into development by reviewing the psychological theories and practices used to create the lessons the chatbot would deliver [31]. In another study, a literature review of the existing health behavior change models was conducted to understand the pros and cons of each model, and to guide the development of a novel behavior change model to structure the chatbot's content [42]. In 1 study, gray literature was sourced through web-based chat threads for a youth helpline, so researchers could better understand content topic preferences and expected answers [40]. Finally, 2 of these 10 studies reviewed the literature to learn more about reward systems and to identify a theoretical basis for chatbot development [35,43].

Patient Engagement and Public Involvement

Overall, the reporting of patient engagement in our included studies was limited making an assessment of PPI using the GRIPP2 challenging. Though 8 studies in our review reported involving patients, 5 provided inadequate detail, making assessing patient involvement impossible [28,29,31,42,43]. Specifically, these studies did not report on the aim of PPI, did not clearly articulate their methods, or did not discuss the role of PPI in their outcomes. The remaining 8 studies were not

evaluated using the GRIPP2 because they did not report development approaches at all [33,34,36] or did not involve patients in the reported approaches [32,35,38,39,41].

Of the 3 studies we assessed using the GRIPP2, 1 study scored 3 points on the GRIPP2 Field [37], with the other 2 scoring 1 point [30,40]. Figueroa et al's study scored 3/5 on the GRIPP2 scale [37]. This study provided a clear description of the methods used for PPI, commenting on how PPI influenced the study and on successful and unsuccessful aspects of the study relating to PPI [37]. This study was also the only one that described 4 different approaches used for development, including co-design workshops, interviews, WoZ, and prototype testing. The authors noted that their co-design sessions "brought unexpected participant preferences and wishes, which were useful in developing subsequent versions" of their chatbot [37]. Further, they recognized the importance of engaging patients in design, testing, and dissemination to develop chatbot interventions that participants would use and benefit from. The remaining 2 studies, 1 by Gabrielli et al [30] and the other by Maenhout et al [40], were each awarded a single point on the GRIPP2 for clearly describing the methods used for PPI. The reporting was such that future researchers could replicate similar development approaches to actively engage patients in research design.

Discussion

Principal Findings

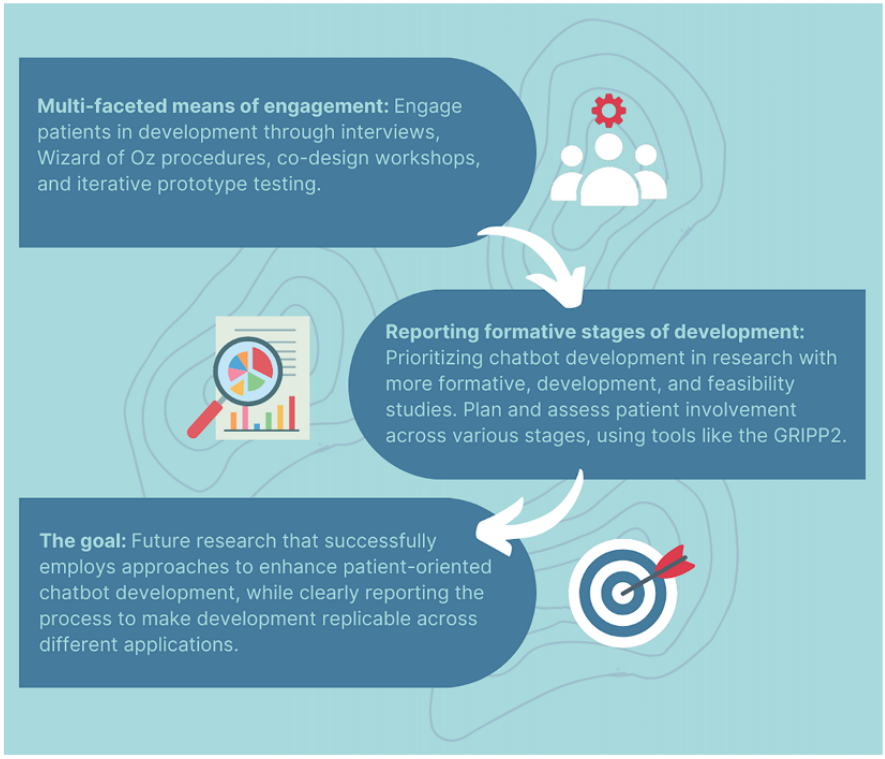
In this review, we described the nontechnical approaches taken for chatbot development and evaluated the extent of patient engagement using the GRIPP2. While promising approaches were shared about the nontechnical steps associated with chatbot development, the level of detail provided was often low, including how patients were involved in the process.

The limited level of detail speaks to the need to prioritize frameworks for implementing digital health tools [44,45]. This will involve a focus on increased formative, development, and feasibility studies and a shift to implementation research that considers embedding and sustaining interventions in context [44,45]. A more detailed focus on the developmental stages and implementation process in research would allow increased replicability of developmental approaches that actively engage patients and progress the field of chatbot research from the end user perspective. An example of this focus on the implementation process includes the formative work conducted by Islam and Chaudhry [46] while developing a chatbot to support the health care needs of patients during the recent COVID-19 pandemic. Their work is an example of detailed documentation of a replicable multi-phased chatbot design study, offering guidance for future research in this area [46]. Additional focus on implementation will ensure the production and monitoring of chatbots that provide quality care and service to patients across short- and long-term timelines [44]. This strategic planning also holds promise to better respond to the requirements of diverse user cohorts, especially those with lower levels of digital health literacy [47].

Although an attempt was made to evaluate the extent of the patient engagement process by the GRIPP2 patient engagement checklist, due to limited detail of reporting, this was only possible in 3 studies [30,37,40]. Many digital health solutions are plagued with low uptake and poor usability as they were developed with minimal patient involvement [48]. As user-centered design and patient engagement are known to improve the quality of research, using engagement approaches throughout the research continuum could result in the identification of system requirements that would be otherwise missed, as well as result in a better understanding of patient needs, higher intervention engagement, and increased intervention effectiveness [49]. Some of the approaches we have identified in this review, including co-design workshops, the WoZ approach, patient interviews, and iterative prototype testing, represent ways researchers can creatively and actively engage patients throughout the development process. Co-design workshops foster a richer understanding of what patients "know, feel, and even dream" [50]. The WoZ approach is a widely accepted evaluation and prototyping methodology for developing human-computer interaction technology [51]. Engaging patients in iterative prototyping and user testing cycles has proven to improve the ease of use and adoption of these interventions [52]. In alignment with the literature, we recommend that researchers taking on health chatbot development projects consider adopting approaches such as co-design workshops, interviews, WoZ, and prototype testing.

Despite the available evidence supporting the benefit of patient involvement in intervention development, there are reasons why approaches that do not directly or actively involve patients may be more appealing to researchers. This notably includes challenges associated with recruitment, particularly when trying to avoid recruitment bias, and the time and resource intensity associated with the overall process [20]. The scarcity of patient involvement may also be related to an underappreciation of the potential benefits of patient involvement in digital health research and a limited understanding of how best to get patients involved [20]. Researchers and practitioners should be aware that there are many different approaches, strategies, and models to engaging patients in chatbot development. We have summarized some approaches in this review, and resources such as the Strategy for Patient-Oriented Research patient engagement framework and the patient engagement in research plan offer practical information to guide patient involvement in the development process [53,54]. Patients can participate at all stages, helping to define health care problems, identify solutions, participate as co-designers of an intervention, and refine the evaluation process [19]. Figure 2 offers the direction in informing future research in patient-oriented chatbot development for lifestyle and wellness interventions, including the application of multifaceted means of patient engagement, use, and thorough documentation of approaches to enhance chatbot development, and clear and replicable reporting of the formative stages of development.

Figure 2. Informing areas of future research in patient-oriented chatbot development for lifestyle and wellness interventions. GRIPP2: Guidance for Reporting Involvement of Patients and Public.



Strengths

We searched 9 of the most relevant bibliographic databases for medical and technology research for this review. No restrictions were placed on the year of publication, country of publication, journal, or study setting. Our study team consisted of multidisciplinary research and health care professionals with relevant expertise who provided direction at each review phase. This review was guided using an established framework proposed by Arksey and O'Malley [21].

Limitations

This review focused on simple voice- or text-based chatbots that engaged in 2-way communication with human users. This led to the exclusion of other forms of conversational agent technology (ie, embodied conversation agents, humanoid and social robots, wearable technology, IoT, virtual avatars, interactive voice assistants, etc) that may have resulted in the finding of additional development and engagement approaches that were not covered in our review. Our review excluded literature from conference proceedings, protocol papers, and other papers lacking an intervention. Moreover, although our proportionate agreement was 0.967 at the title and abstract screening stage, there was only "fair" agreement between reviewers (Cohen κ=0.309). This "fair" agreement between researchers highlights the challenges in reviewing a heterogeneous body of literature. With ongoing meetings and refinement of our inclusion and exclusion criteria, the Cohen κ statistic improved to an "almost perfect" agreement at the full-text review stage (Cohen κ=0.843). Additionally, due to the limited detail available within the included studies, our team could not conclusively assess patient involvement in chatbot development; greater attention to reporting patient involvement in chatbot development and testing in future research will help with this limitation. Finally, we acknowledge that scoping reviews have numerous shortcomings, including limitations of rigor and potential bias stemming from the absence of a quality assessment, among others [55]. However, the literature on chatbot technology remains highly heterogeneous at this time, and scoping review provided a systematic method to map the current state of the literature.

Conclusion

In conclusion, this review provides a menu of options that can be used for the nontechnical steps associated with chatbot development for interventions supporting lifestyle and wellness interventions. The identified study limitations hold promise to guide the inclusion of patient engagement and the improved documentation of the engagement and development of chatbots in future health care interventions. Given the importance of end user involvement in the development of digital technology, we hope that future research on chatbot development will take the opportunity to carry out a more systematic reporting of the chatbot development and implementation process and will actively engage patients as key members of the codevelopment process.

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Conflicts of Interest

None declared.

Multimedia Appendix 1
Search strategy documentation.
[PDF File (Adobe PDF File), 81 KB - jopm_v15i1e45772_app1.pdf]

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Abbreviations

AI: artificial intelligence
DHI: digital health intervention
GRIPP2: Guidance for Reporting Involvement of Patients and Public
IoT: Internet of Things
PPI: patient and public involvement
PROSPERO: International Prospective Register of Systematic Reviews
RCT: randomized controlled trial
WoZ: Wizard of Oz

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Extraordinary Lives

An Extraordinary Voice Expressed Through Humor: A Tribute to Casey Quinlan

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Abstract

The *Journal of Participatory Medicine* introduces *Extraordinary Lives*, a new journal section celebrating the voices and work of steadfast advocates of participatory medicine that we have lost. This inaugural essay spotlights Casey Quinlan, a patient activist who effectively used her humor and incisive analysis of health care to encourage others to strive for meaningful change. A first-generation "professional patient," Casey served as a role model who inspired many to share their stories and achieve genuine partnerships in care delivery. A maker of "good trouble," her voice and stance were part of her power and influence in disrupting the status quo. We present her fight for personal access to health data, her aspiration for personally customized evidence, and her drive for all people to control their health and their health care.

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KEYWORDS

participatory medicine; co-design; co-production; patient engagement; patient empowerment; electronic health record; patient portal; open notes; evidence-based medicine; shared decision-making

Introduction

The *Journal of Participatory Medicine* is introducing a new journal section called *Extraordinary Lives.* Papers in this category are essays celebrating and commemorating advocates and pioneers the world has lost, yet who have left a lasting impact on participatory medicine. These essays lift a torch to champions who contributed to transitioning health care from a top-down, revenue-driven enterprise [1] to one exhibiting genuine partnership between patients and professionals delivering care.

Our journal's mission is to generate knowledge on co-design to improve health care and research, and to demonstrate how the internet and digital services enable people to achieve healthy lives [2]. Our field is dependent upon and indebted to advocates who contribute to these aims. To inaugurate *Extraordinary Lives*, we highlight the life and work of Casey Quinlan (1952-2023). Many have written about her wisdom, insights, and contributions [3,4]. Through her unquenchable activism targeted across many participatory care domains, Casey is an exemplar to launch such a tribute. Here, we describe only a few of her numerous contributions; we focus attention on her efforts in advancing personal access to health data, activating patients to fully participate in their care, and encouraging patients and caregivers to contribute their expertise across the health care ecosystem.

Performer, Journalist, Blogger, Podcaster, and Activist

Casey was born Mary Martha Casey at the US Naval Academy in Annapolis, Maryland, to Martin Michael Casey and Marie Elizabeth Rodgers Casey. Her father was a Navy captain and

her grandfather was a Navy rear admiral; unsurprisingly, Casey considered herself a warrior throughout her life. Early on, she dropped Mary as her first name and substituted Casey, subsequently taking Quinlan as her last name in honor of her paternal grandmother.

Raised on both the west and east coasts of the United States and in England, she gained a broad worldview that helped shape a skeptical perspective on institutions. In grade school, she adopted the moniker "Mighty Casey" after being teased on the playground about "Casey at the bat" striking out. Foreshadowing her trademark feistiness, she declared she was, indeed, Mighty Casey, but that *she* did not strike out!

Casey studied theater and performance at the University of San Francisco, then moved to New York to study at the American Conservatory Theater, HB Studios, New York University, and the American Comedy Institute. She performed stand-up comedy at Caroline's, Gotham Comedy Club, Catch A Rising Star, and the New York Comedy Club. Pivoting to broadcast news and sports, she worked as a field producer and engineer for NBC News, where she covered stories for Dateline and Today, political campaigns, wars, NFL Playoff games, Stanley Cup hockey, and the NBA.

Quinlan later moved to Virginia, where she started Quinlan Media Services. She subsequently became VP of Marketing and Operations for Skywire Uplink and launched Mighty Casey Media, LLC. She was awarded the ABWA Richmond Region Business Woman of the Year in 2006 and the Toastmasters International Distinguished Toastmaster Award in 2007. The following year, she was diagnosed with breast cancer. After her treatment, she devoted her business and personal energies to health care advocacy and never looked back.

In 2009, she published a book about her experience, *Cancer for Christmas: Making the Most of a Daunting Gift* [5]. A description of the book on the Amazon website reads [6]:

> *"Whoopee – cancer!" That's not your average reaction to a cancer diagnosis, and Casey Quinlan isn't your average patient. When, after her 15th mammogram, she won the booby prize – breast cancer – her first reaction, after downing a stiff drink, was to cover her own cancer story with the same relentless inquiry she brought to her career in network television news, and that informs her work as a 'business storyteller' and branding consultant. Casey's approach to treatment: be an active participant, not a passive consumer. Her metaphor for managing treatment? "It's like a car wash. When you go to a car wash, do you want to be inside the car, or strapped to the hood? Ask questions, make sure you understand the answers – you get to stay inside the car. Otherwise, you get lots of soap and wax up your nose!"*

Active Participation in Care

Casey was a thought leader, speaker, and all-around maker of "good trouble." She was armed with the knowledge—accrued over decades by academics and quality improvement experts—that the *power of patients* could transform and improve health care quality, efficiency, and effectiveness, and lead to better outcomes and patient satisfaction [7-9].

She espoused the principles of participatory medicine, co-design and co-production, where stakeholders do not merely recognize but embrace patients' and caregivers' contributions. Riding the wave of the digital era and the democratization of health data, Casey implored people to study their conditions and treatments, examine clinician quality, and engage peers to learn about their experiences. She gained considerable experience in how patient contributions produced greater autonomy and sense of control. She published the following on the Society for Participatory Medicine's e-Patients Blog [10]:

> *Patient means different things, to different people, at different times. Whatever anyone's view of being a patient is, we all have one goal: that others on my care team and around me will respect my definition of my status and seek to understand what it means to me. It can be a role that comes and goes, and returns, different than before, or similar. It can be part of my identity – something felt and lived strongly or coexisting quietly. This too, can develop and change. It can be a view others have toward me, whether I share their view or not. It can mean I'm highly dependent on others (I'm anesthetized for surgery) or highly independent (I'm self-managing) or co-dependent (we're co-managing).*

While a collaborative approach fosters trust and mutual respect, Casey keenly understood that patient-clinician interactions are a tricky dance. She wanted patients to challenge health professionals to be meaningful partners in their care and to gain power in medical decision-making. She expressed the following in her book [5]:

> *I think that, over the centuries, while medicine has been viewed as a calling, some doctors have misunderstood their place in the doctor-patient relationship. We're partners in our care. People – again - the ones called "patients" in this party, are not worshipers at the altar of medical professional knowledge, nor are we lesser beings because we don't have MD behind our names. I suggest that each and every doctor on the planet invest in some communication training, for themselves and their staff. Remember, you're the partner in your own care.*

In December 2014, Consumer Reports posted an article [11], "The surprising way to stay safe in the hospital," summarizing the results of a survey of 1200 people who had been recently hospitalized. Patients who said they received respectful treatment from clinicians also reported fewer medical errors and better experiences during their hospital stays. Considering this report, Casey blogged [12]:

> *Engagement may be the buzzword, but accountability is the watchword, for both clinical teams and patients. We all have to participate. Which we can only do if we're fully informed. There is a good and a bad way of challenging your doctor. The notion that 'you are the expert when it comes to your body and the doctor*

> *is the expert when it comes to medicine' is a good rule of thumb. There should be a spirit of teamwork that includes shared observations, knowledge and information and asking questions – but not making accusations.*

Getting Your Data and Customizing Evidence

Information is power, and Casey exhibited unparalleled demand for patients and caregivers to have ready access to their electronic health record data. With courage and creativity, she tattooed a QR code on her upper chest which, for a while, opened digital access to her personal record data [13]. She first posted about this in 2015 [14]:

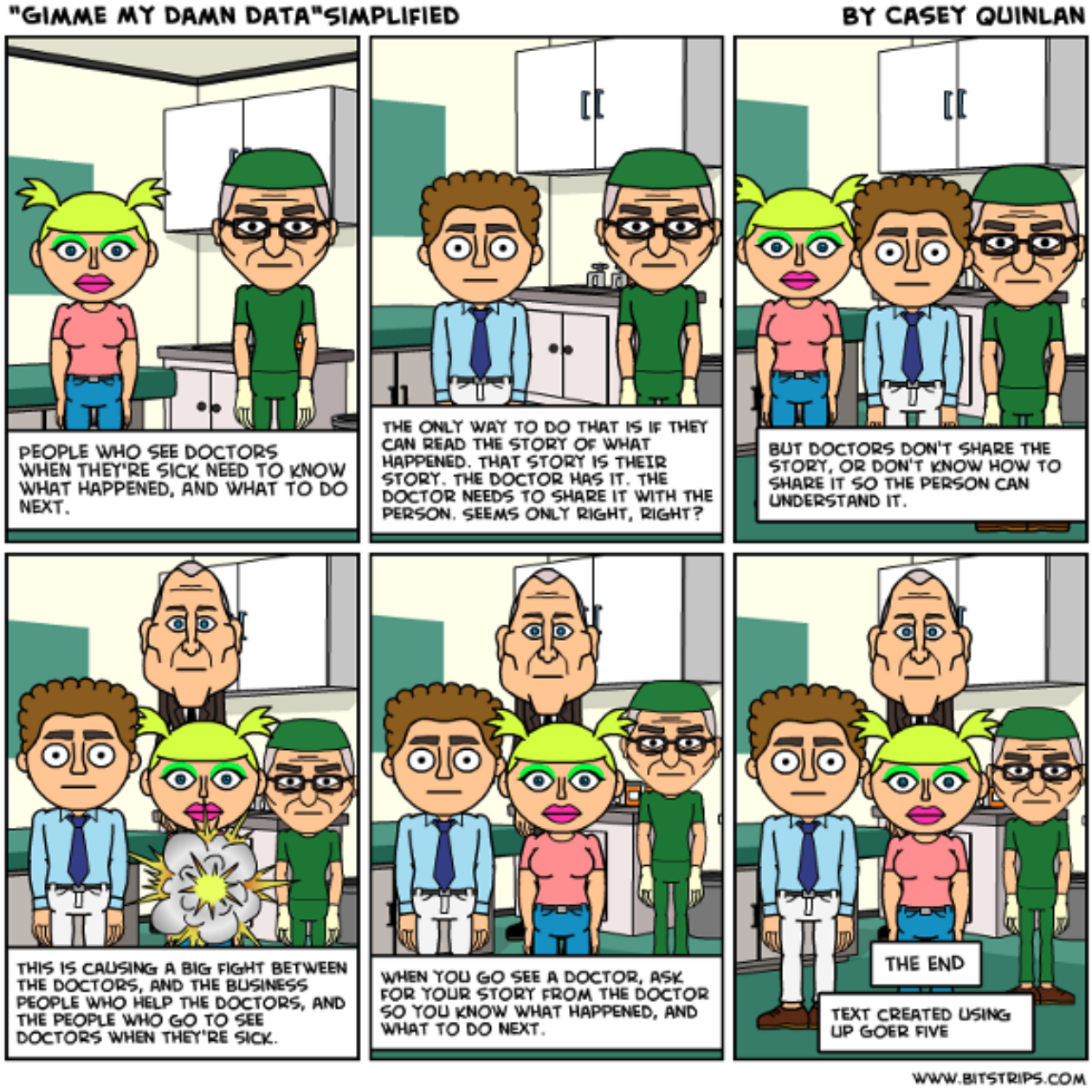
> *Why did I do this? Because I've been waiting for the medical-industrial complex to deliver on their promise of health information exchange (HIE), the promise that they've been making for years, but have yet to fork over. I can, and do, securely move money around the globe at the click of a mouse. I do it via bank accounts, purchase agreements, contracts with clients. Most people do. But my healthcare record — which is MINE, as much as it is the property of the medical providers who gave the care it describes — is in fractured bits and pieces all over everywhere.*

As a patient activist, Casey used all communication channels to implore patients to access their full health records, including cartoons demonstrating this plea (Figure 1). She wrestled with professionals disparaging patients who used the internet, often referred to as "Dr Google" [15]. She argued that well-informed people ask better questions and choose treatments aligned with their values and preferences. Yet, she also knew publicly sourced information could be unreliable and appreciated that searching may produce anxiety or delay seeking professional advice. She was passionate about reputable, evidence-based knowledge generation, and contributed to the efforts of the Cochrane Collaboration, a robust source of high-quality systematic reviews. She actively promoted the Cochrane Consumer Network, a network anyone interested in high-quality evidence can join [16]. She wrote [17]:

> *Cochrane popped up on my radar screen sometime in the last decade or so, during the time I was scrambling to get on top of managing my parents' care in the last few years of their lives. It came in handy as I was sifting through my decision tree during cancer treatment ten years ago, and as I've become more and more interested in killing off quackery and over-, under-, and mis-treatment in medicine in my work as a citizen science activist and ground-level health policy wonk. We're all in this together, and Cochrane can help us move the needle toward what I call "Goldilocks medicine" – the right treatment for the right patient, at the right time – at a faster rate.*

She was invited to speak at the 2018 Cochrane Collaboration international meeting and coauthored a paper that promoted consumer access to trustworthy evidence customized to patient preferences and contexts [18]. The authors reasoned that "the audience for high quality evidence is much wider than merely health care professionals – and that there is a case to be made for creating tools that translate existing evidence into tools to help patients and clinicians work together to decide next steps." From this work, Casey continued to argue for greater public access to research locked behind "paywalls" and for patients and families to provide more genuine and robust contributions to decision support tools.

Figure 1. "Gimme My Damn Data - Simplified," a cartoon by Casey Quinlan (reproduced from Mighty Casey Media [19], which is published under Creative Commons 4.0 Non-Commercial License [20]).



Patients Are Experts: Ignore Them at Your Peril!

Casey's trademark humor and incisive analysis of the health care system made her a highly sought-after speaker and consultant. She was honored with the Right Care Alliance Leadership Award (2017) and the WEGO Health Best One Liner Award. She served on multiple boards and steering committees, notably the Society for Participatory Medicine, Health Datapalooza, and the Light Collective. She worked tirelessly to raise awareness about the needs and rights of all patients and particularly encouraged metastatic breast cancer research. She served as an expert patient and exemplar of co-design at health technology events and passionately helped drive patient and consumer attendance at health industry and scientific conferences [10]:

> *If you're planning a healthcare industry event that is focused on patient engagement, patient-centered design, patient-centered care, patient-centered technology, or touches on patient care in any part of the healthcare setting or system, you have to include patients on your program or be judged Patients Excluded. Nothing about us without us.*

Casey advocated for compensating patients and caregivers for their time spent informing health care system improvement, whether on advisory boards, speaking engagements, or providing feedback, remarking that "warm handshakes and cold bagels" were insulting. Instead, Casey advocated for fair payment models for patients and caregivers—who she called *ground level experts*.

Casey also felt that all revenue generated from monetizing patients' personal data should be shared with the people who are, after all, she reasoned, the source of such data. In a blog post, she elaborated [21]:

> *Let's take to the streets, the halls of Congress and state capitals, policy meetings, and star-chambers from sea to shining sea, to demand compensation for the ginormous wads of cash that are minted—literally—from our bodies, bones, blood.*

A Unique Role Model

Casey's outspoken nature could be intimidating until one got to know her. She took time to listen. She supported and encouraged many others to find their voices and tell their stories. Her courage and determination left an indelible legacy of advocacy for a more equitable and effective health care system. She approached her cancer as intensely as she lived her life. Friends, colleagues, and loved ones found solace in the fact that her principles and values remained evident even at the end of her life. During a Health Hats podcast interview [22], she retorted:

> *So, f**k cancer, I'm not done, and I'm not quitting until I'm dead. And then I want you all to carry me off the battlefield on my shield and then keep fighting. Because that's the only way we're going to hack this universe into a more human-friendly place.*

Casey was a role model who gave us the courage to speak up, for ourselves and for all those affected by bureaucracy, inequity, and arrogance. Today, as artificial intelligence (AI) garners much attention, we can imagine Casey front and center, promoting the promise of AI and large language models to sharpen patients' engagement with decision-making, while cautioning us all about its risks. Her irreverence, prominently displayed in the name of her podcast, Healthcare is Hilarious!, admonished us to embrace joy as we figure it all out [5]:

> *My philosophy in a nutshell: Life is 100% fatal. Let's have fun while we're here.*

Conflicts of Interest

SSW and MFH are editors-in-chief of the *Journal of Participatory Medicine*. DvL is an associate editor of the *Journal of Participatory Medicine*.

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Abbreviations

AI: artificial intelligence

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Original Paper

Coproduction of Low-Barrier Hepatitis C Virus and HIV Care for People Who Use Drugs in a Rural Community: Brief Qualitative Report

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Abstract

Background: People who inject drugs are experiencing syndemic conditions with increasing risk of infection with hepatitis C (HCV) and HIV. However, rates of accessing HCV and HIV testing and treatment among people who inject drugs are low for various reasons, including the criminalization of drug use, which leads to a focus on treating drug use rather than caring for drug users. For many people who inject drugs, health care becomes a form of structural violence, resulting in traumatic experiences, fear of police violence, unmet needs, and avoidance of medical care. There is a clear need for novel approaches to health care delivery for people who inject drugs.

Objective: This study aimed to analyze the process of a multidisciplinary team—encompassing health care professionals, community representatives, researchers, and people with lived experience using drugs—that was formed to develop a deep understanding of the experiences of people who inject drugs and local ecosystem opportunities and constraints to inform the cocreation of low-barrier, innovative HCV or HIV care in a rural community. Given the need for innovative approaches to redesigning health care, we sought to identify challenges and tensions encountered in this process and strategies for overcoming these challenges.

Methods: Analysis was based on an in-depth review of meeting notes from the project year, followed by member-checking with the project team to revise and expand upon the challenges encountered and strategies identified to navigate these challenges.

Results: Challenges and tensions included: scoping the project, setting the pace and urgency of the work, adapting to web-based work, navigating ethics and practice of payment, defining success, and situating the project for sustainability. Strategies to navigate these challenges included: dedicated effort to building personal and meaningful connections, fostering mutual respect, identifying common ground to make shared decisions, and redefining successes.

Conclusions: While cocreated care presents challenges, the resulting program is strengthened by challenging assumptions and carefully considering various perspectives to think creatively and productively about solutions.

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KEYWORDS

hepatitis C; HIV; coproducing care; testing; people who use drugs; HCV; rural community

Introduction

Hepatitis C Virus and HIV Among People Who Inject Drugs—Treatment Challenges

Infections such as hepatitis C (HCV) and HIV disproportionally affect people who inject drugs. Globally, over half of people who inject drugs are infected with HCV, and nearly 1 in 5 with HIV [1-5]. Many people who inject drugs are unaware of their HCV or HIV status, and rates of accessing treatment are low. For instance, fewer than 10% of those infected with HCV receive treatment [6], increasing morbidity and contributing to ongoing virus spread [7].

Several factors contribute to the low rates at which people who inject drugs access treatment. People who inject drugs are often stereotyped and treated poorly in professional settings; many have histories of traumatizing experiences with medical care [8,9]. These experiences cause health care mistrust and fear of being criminalized for drug use rather than treated for health needs [10]. Differences between the values and goals of clinicians and people who inject drugs can exacerbate difficulties between groups [11]. Medical professionals' focus on achieving abstinence from drug use leads to missed opportunities for people who inject drugs to engage in health care if people who inject drugs are not interested or able to stop using drugs. Treatment for HCV is not always made available to people with active drug use. Some service providers exclude people who inject drugs from HCV treatment due to concerns about poor treatment adherence and risk for reinfection, despite national guidelines explicitly recommending the inclusion of people who inject drugs in HCV treatment [12,13]. Even in places without treatment restrictions, myths and knowledge gaps may limit treatment access [14-17]. Further, even when individuals are aware that HCV is curable, people who inject drugs may not prioritize treatment for an infection that is asymptomatic and not immediately life-threatening over more pressing needs associated with chaotic drug use. HCV treatment may also be less appealing for those who witnessed individuals taking first-generation HCV medications that had profound side effects [18].

In rural areas, compounded barriers affect treatment access, including long distances to care and less health care availability, lack of transportation, limited internet and phone connectivity, lower socioeconomic status and associated concerns regarding medication costs, and unstable housing [19-21]. These same factors may also help explain why people who inject drugs in rural areas receive HCV or HIV testing less frequently than their urban counterparts [22]. Barriers to treatment in rural areas coexist with increasing opioid use and opioid-related mortality rates [23].

The Value of Coproducing Care

Extensive research highlights that coproducing care and programs increases the likelihood that the resulting programs match the needs of the target communities, resulting in better health outcomes, patient satisfaction, and even cost savings [24-26]. Principles of community engagement highlight the importance of fostering relationships, building off existing trust, being flexible and responsive, promoting ongoing engagement through mutual understanding, user-centeredness, and reciprocity, empowering community members, being willing to be questioned and challenged, creating a safe and supportive environment, and respecting differences [27]. Following these aforementioned principles and providing opportunities for individuals with lived experience of drug use to have longitudinal roles within the project helps to avoid tokenism and promote genuine engagement [28].

Innovating HCV or HIV Care Delivery

With 1 year of funding from a health care delivery innovation laboratory [29], we used a human-centered design approach to develop a program to facilitate the connection between health care providers and populations at risk for sex- and drug-related harms [30,31]. We assembled a multidisciplinary team of health care professionals, community representatives, researchers, and people who inject drugs. We invited health care professionals with a wide range of expertise in infectious diseases and addiction: an infectious diseases and addiction medicine physician, an HIV nurse care manager, an HIV outreach nurse, and a psychologist embedded in the HIV program with a focus on trauma and addiction. To augment clinical perspectives, the team included a medical anthropologist with experience working with rural communities as well as several harm reduction specialists with lived experience of drug use who work for community-based organizations serving people who struggle with addiction, those are, a syringe service program and an addiction treatment program. The team also included a person with lived experience of drug use as a patient innovation partner; the patient innovation partner role was designed by the health care delivery innovation laboratory to ensure teams included an embedded team member who was encouraged to share the patient perspective throughout the design journey. In composing the team, we aimed to include multiple perspectives from within as well as from outside the health care system. Due to COVID-19 and social distancing recommendations, project team meetings occurred digitally via Zoom (Zoom Video Communications).

To be responsive to the needs and preferences of people who inject drugs and to ensure a range of perspectives and experiences with drug use and health care informed the program design, the team developed a community advisory board (CAB). Most CAB members were current or former clients of the syringe service program, and one was an employee of another syringe service program. All 8 CAB members had a history of injection drug use and 4 were currently using. While not all CAB members shared their HCV or HIV status, 3 indicated prior treatment for HCV and one indicated being actively treated for HIV. The CAB included 4 men and 4 women and, consistent with area demographics, was mostly White (7 White and 1 Black). CAB members lived all over the rural northeast,

including New Hampshire, Vermont, Maine, and upstate New York.

The CAB met with members of the project team via the web, once or twice per month for 6 months, providing feedback and generating ideas regarding educational and promotional materials, testing and referral processes, and outreach strategies. CAB members were also given opportunities to participate in individual interviews to share additional program development ideas and suggestions.

A notable contribution of the CAB was the development of the name for the program; they came up with "To the Point," which resulted from a conversation around sticking to the goals of the program and a play on words when considering the point of a needle transmitting virus. "To the Point" was preferred to the project's working name, "Connect to Cure," as curing HCV or HIV is often not of immediate interest—especially when infection status is not yet known—among people who inject drugs.

This work resulted in a new care pathway grounded in principles of trauma-informed care and harm reduction that is embedded within an existing syringe service program. The program developed is a novel community-based, peer-led, HCV or HIV testing service in rural Vermont and New Hampshire. The program is responsive to the schedules and preferred testing locations of people who inject drugs and relies on staff already embedded within the community with lived experience of drug use and established trust. During testing encounters, clients are offered immediate, digital connections to medical providers and are given harm reduction information and supplies including naloxone, safe injection equipment, wound care supplies, and information about HCV and HIV.

While the project team was deeply committed to designing a program responsive to community preferences and needs, the process of coproduction was fraught with difficulties. The purpose of this paper is to shed light on the challenges this multidisciplinary team encountered along the way in coproducing this new care model and the strategies used to navigate these challenges.

Methods

Overview

A general inductive approach was used to analyze the data, whereby instances of challenge or tension were used to identify broader categories of challenge, without any preconceived notions or theories guiding the analysis [32]. While throughout the project period team members had informally identified various challenges and tensions throughout the process, at the conclusion of the 12 months of the project, the first author carefully reviewed all notes from the project year. Notes included weekly meeting notes from throughout the project, project-related emails, and monthly CAB meeting notes. For all notes, excerpts that reflected differing perspectives, tensions, or uncertainty regarding how best to proceed with program design were pulled. These instances were then sorted into thematic areas of tension or challenge, with thematic categories refined iteratively, as additional data were reviewed. The resulting challenge areas were brought back to the full project team on multiple occurrences—both in writing and via verbal discussion—as a form of member-checking to review for accuracy, thoroughness, and appropriate categorization. Because the analysis focused on challenges encountered by the project's multidisciplinary team throughout the project year, CAB members were not part of this member-checking process.

Ethical Considerations

The team's work was reviewed by Dartmouth Health's institutional review board and determined to be quality improvement, not human participants' research. All project team members were compensated for their time on project-related activities, including this analysis, either through effort brought out through their institution or through payment for time, depending on their work situation. CAB members were sent a US $25 gift card for their participation in each meeting. In addition, clients were incentivized with US $25 gift cards for participating in the HCV or HIV testing and subsequent steps in the care cascade (ie, follow-up blood work, a clinical visit, initiating medications, finishing medication, and final blood work).

Results

In total, 6 areas of challenge in coproducing care were identified: scoping the project, pace and urgency, adapting to web-based work, navigating the ethics and process of fair payment, defining success, and situating for sustainability. Each of these is discussed below.

Scoping the Project

When this project launched, the focus—originally defined by the project lead, an infectious disease physician—was on helping individuals who already knew they were HCV-infected to follow through on receiving treatment and cure. However, through the design thinking process, it became clear that this focus was too downstream and was missing the larger need to help people in the community learn their infection status, and many were hesitant to engage with health care. Accordingly, the project's focus shifted to increasing HCV or HIV testing and linkages to health care. Social injustices and fundamental public health problems were identified as the project progressed. While the team recognized that testing for HCV or HIV was limited, other more immediate medical needs such as wound care, management of acute infections, and mental health treatment were identified as unaddressed. Further, basic needs for food and shelter were unmet, and access to phone service, stable internet, and transportation remained challenging. Several team members wanted to tackle these broader health and social challenges—and tension emerged between staying focused on project goals and keeping these issues front of mind. Academia encourages specific aims that are both measurable and achievable; scope creep and losing focus can compromise traditional notions of success or measurable progress. Accordingly, the team maintained the HCV or HIV testing focus, acknowledging that the team's composition and time constraints were not aligned for addressing these broader public health issues, but integrated an awareness of social determinants of health and health inequalities into the program design. We managed these tensions

by framing the initial work as a proof-of-concept pilot, being explicit about the program's current limits, but maintaining the goal of future expansion to address greater, more immediate needs.

Pace and Urgency

Health care quality improvement is often slow, with various processes and approvals required for changes [33], but developing procedures to ensure safety and confidentiality is critical. Moving slowly to ensure adequate data are collected to evaluate program effectiveness to support publications and grants enables broader impact potential but typically does not have immediate benefit. For those immersed in the community faced with people suffering daily, there is an urgent need for change and frustration with the slow, bureaucratic processes to make health care changes. The low tolerance for risk and error in health care is at odds with the substantial and urgent health needs that are not being addressed for people who inject drugs. Accordingly, there was tension between the clinical and research members of the team's desire to pause to develop strategies to ensure safety, confidentiality, and rigorous data collection and the community representatives' and people who inject drugs' desire to bring testing into the community and offer support as soon as possible. This tension over urgency is also evident in writing about the work; peer-reviewed academic publications tend to encourage a neutral voice, but a neutral tone may mask the emotion involved and the urgent need for change.

Adapting to Web-Based Work

The project team's work occurred almost exclusively via the web. Interacting remotely makes it challenging to develop the personal connections that bond teams and build trust and belonging [34]. Special efforts to connect were necessary, such as: dedicating time to empathize with personal struggles and celebrate successes, creating opportunities to engage in occasional, in-person meet-ups, and recognizing the passion that team members brought to the work. These explicit efforts to strengthen relationships and build trust are essential to successful coproduction [35]. In addition, the team actively attempted to reduce hierarchies in team structure and minimize power differentials, striving to build consensus by seeking out and listening to all team members, and fostering mutual respect in the search for common ground. In the absence of common ground, individuals may abandon the work, reinforcing existing cultural divides. Accordingly, the search for common ground requires an ongoing, continued, and explicit effort.

Navigating Ethics and Practice of Fair Payment

The team had to navigate the ethics of fairly compensating people who inject drugs for sharing their experiences to inform program design—both through the CAB and initial design thinking interviews—while maintaining a noncoercive relationship and abiding by prohibitive institutional rules regarding hiring and payment. Prior research suggests that compensation can build trust and demonstrate reciprocity and respect, enabling engagement to evolve [36]. While there may be concern about people who inject drugs redirecting cash toward drug use, research does not validate this concern [37] and questions the ethics of "item restrictions" in financial support programs (eg, food assistance). Institutional rules prevented employing CAB members in the health system due to a required drug test. Other compensation options relied on quarterly payments—a long wait for individuals with minimal financial resources. These limitations can prevent people who inject drugs from engaging in such projects. Navigating these payment challenges also raised questions about the role and ethical responsibility of large, well-funded institutions to their surrounding communities. Ultimately, the team chose to use cash gift cards to balance regulation with autonomy and respect for privacy.

Redefining Success

After the initial month of testing, no one who tested positive for HCV had connected to health care, despite efforts to provide low-barrier engagement opportunities. Several team members saw this as a failure; others highlighted that more people knowing their viral status was itself a success and empowering individuals to take the next step in the care cascade, when and if they decide to, was essential. Recognizing the pervasive distrust of the medical community, the team redefined notions of success. Accordingly, a positive, nonjudgmental, clinical experience may hold value in and of itself [38]. The team viewed these as corrective emotional experiences; when individuals engaged in medical-like encounters without the stigma they have previously experienced, future health care receptivity may increase. The team also acknowledged that individuals may move through this care cascade at different speeds—and adjusted expectations around the immediacy of follow-up. The project's time constraints encouraged rapid measurement, yet people who inject drugs may take months to take the next step from testing to treatment. By month 2 of the program, the program began to see some connections to health care.

Situating for Sustainability

The goal of the health care innovation laboratory is for new care pathways to be sustainable after the initial year of funding. Sustainability is vital so that gains in community trust or headway in overcoming barriers to health care are preserved. Return on investment calculations can justify new positions for initiatives exclusively situated within the health care system. This project, however, does not immediately translate into resource savings or new revenue generation for the health care institution. While providing compassionate linkages from the community into the health care setting may introduce new patients into the system or encourage earlier care for infections and wounds rather than costly emergency visits, potential revenue and cost savings are delayed. Accordingly, the team was challenged with advocating for and obtaining ongoing financial support from the health care system while operating the program outside of the traditional walls of the institution.

Discussion

This coproduction process revealed several insights that may help others planning to engage in similar partnerships. Committed partnerships enable teams to move the needle with respect to care; however, even with dedication, creating change remains difficult. Building bridges to overcome cultural barriers, by engaging people who inject drugs in the team and program

design and including team members with experience both as people who inject drugs and in service delivery, helped develop a shared understanding of challenges, for example, people who inject drugs' experience of health care trauma and mistrust. By acknowledging deep-seated challenges, and validating and acknowledging the values of people who inject drugs, the resulting program design was sensitive to community needs and health care limitations.

The importance of ongoing open conversation and support is not to be understated. There may be a frankness or directness that emerges that is uncomfortable, but this honesty should be recognized and appreciated as an opportunity to address personal frustrations and strengthen relationships [39,40]. Refocusing on a shared mission, that defies traditional ideas around working with people who inject drugs that views stopping substance use as a primary success indicator, and acknowledging and celebrating small successes, helps build connection and is at least a step toward re-energizing and countering the burnout frequently experienced by individuals working in this area [41]. These partnerships benefit from individuals entering the work with mental flexibility and a willingness to challenge assumptions and think creatively and productively about solutions; the design thinking approach used for this program's development encouraged such innovative thinking [42].

This work also highlights the important role that community partnerships can play in sustainability, leveraging each group's strengths to support common goals and shared missions to help the communities they serve. This work is currently being sustained through a combination of health center staff investment, syringe service program staff support, and philanthropic funding. These partnerships were possible due to strategic and comprehensive communication with health care and community organization leaders, creating shared buy-in, support, and ownership for the project's ongoing success [43]. By distributing the work and costs of running the program, and by clearly communicating the need for change, program goals, and the work needed to achieve those goals, resistance to change is reduced [44]. These collaborations will likely evolve over time, as incentives, constraints, and priorities shift. Efforts to evaluate program impact on clients, providers, and the communities in which this work occurs, will hopefully facilitate ongoing investment from all parties [45,46]. The ongoing work also requires shared responsibility and flexibility on the part of the health system, to incorporate care innovations that disrupt usual practices. Fostering a willingness to work through ambiguity can help establish an institutional context that can accommodate change.

There are several limitations to acknowledge in this analysis. First, since meetings were not audio-recorded, challenges identified in the coproduction process may have been overlooked if they were not reflected in the team meeting notes or actively recalled by team members. Consequently, it is possible that additional challenges were encountered that are not reflected in the discussion above. Further, because the notes were taken by a member of the innovation laboratory staff, it is possible the academic lens through which notes were taken may have led to some challenges being overlooked. However, the diversity of the team, including an anthropologist sensitive to team dynamics, and the iterative discussions of findings hopefully minimized potential omissions. In addition, while this analysis focuses on the coproduction process among the project team members, future research could also examine challenges in the context of the CAB. Despite these limitations, this analysis demonstrates that several tensions occur throughout the coproduction process. The negotiations and thoughtful considerations of various perspectives that emerged from these tensions supported the development of a program that is sensitive to the preferences and needs of the population it seeks to serve. Future work will evaluate the initial outcomes of this program.

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Conflicts of Interest

None declared.

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Abbreviations

CAB: community advisory board
HCV: hepatitis C virus

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Original Paper

An Ethics Action Plan for Rare Disease Care: Participatory Action Research Approach

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Abstract

Background: Owing to their low prevalence, rare diseases are poorly addressed in the scientific literature and clinical practice guidelines. Thus, health care workers are inadequately equipped to provide timely diagnoses, appropriate treatment, and support for these poorly understood conditions. These clinical tribulations are experienced as moral challenges by patients, jeopardizing their life trajectories, dreams, and aspirations.

Objective: This paper presents an ethical action plan for rare disease care and the process underlying its development.

Methods: This action plan was designed through an ethical inquiry conducted by the *Ethics and Rare Diseases Working Group*, which included 3 patient partners, 2 clinician researchers, and 1 representative from Québec's rare disease association.

Results: The plan is structured into 4 components. Component A presents the key moral challenges encountered by patients, which are the lack of knowledge on rare diseases among health care workers, the problematic attitudes that it sometimes elicits, and the distress and powerlessness experienced by patients. Component B emphasizes a vision for patient partnership in rare disease care characterized by open-mindedness, empathy, respect, and support of patient autonomy from health care workers. Component C outlines 2 courses of action prompted by this vision: raising awareness among health care workers and empowering patients to better navigate their care. Component D compares several interventions that could help integrate these 2 courses of action in rare disease care.

Conclusions: Overall, this action plan represents a toolbox that provides a review of multiple possible interventions for policy makers, hospital managers, practitioners, researchers, and patient associations to critically reflect on key moral challenges experienced by patients with rare diseases and ways to mitigate them. This paper also prompts reflection on the values underlying rare disease care, patient experiences, and health care workers' beliefs and behaviors. Health care workers and patients were the primary beneficiaries of this action plan.

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KEYWORDS

community-based participatory research; rare diseases; bioethics; delivery of health care; ethics; clinical; patient participation; empowerment; education; medical; attitude of health personnel; patient education as topic; patient partnership

Introduction

Background

A disease is considered rare when its prevalence is estimated to be 1 in 2000 [1]. Owing to their rarity, rare diseases seldom attract the interest of researchers. As a result, their symptoms, mechanisms of action, and therapeutic avenues are poorly covered in scientific literature and clinical practice guidelines [2]. This lack of knowledge often translates into significant difficulties for patients in obtaining timely diagnoses [2]. The prolonged quest for a conclusive diagnosis, known as the diagnostic odyssey, may last several years [3]. Through this process, patients may be misdiagnosed or undergo several inconclusive consultations by specialists [3,4]. This wait is difficult for patients, especially given the lack of satisfactory explanations for their health struggles, and elicits profound uncertainty [5], distress [6], and frustration [7]. Moreover, receiving proper treatment is often impossible for patients with rare diseases; less than 10% of rare diseases benefit from effective treatment [8]. This limited clinical support exacerbates disease manifestations and fuels additional challenges in daily activities, social interactions and relationships, and professional activities [9]. Such difficulties are common to most rare diseases [10] and may lead to moral challenges.

Moral challenges are experiences that are unacceptable in the eyes of patients because they put their values at stake [11] (Racine E, unpublished data, September 2023) and hold great significance, often having profound and multifaceted implications [12]. In health care, diagnostic and prognostic uncertainties, as well as disability, create moral challenges because they impede the ability of patients to pursue important values. These distressing situations put patients' life trajectories, dreams, and aspirations at stake [13]. Despite the moral distress elicited by these challenges, they have only been superficially investigated and even less so in relation to adults living with rare diseases [14,15]. These salient moral challenges call for ethical inquiries, which involve acquiring an in-depth understanding of moral challenges, critically reflecting on the values they jeopardize, and imagining ways to address them [16,17].

Ethical inquiries are processes by which difficult moral challenges are understood and addressed by discussing possible responses, enacting the most promising response, and evaluating these responses [18]. In other words, ethics and ethical inquiries take moral challenges as objects to ask questions about their nature, value, meaning, and impact in terms of broader notions of fulfillment, such as human development and flourishing. This means that when an experience is designated as being morally problematic (eg, a patient with a rare disease experiencing a situation as a challenge to their own self-esteem or autonomy), this signifies that this experience is lived and experienced as a challenge with respect to one's values and self-concept, thus calling for a response. Moral experiences are anchored in daily life, including the challenges faced by patients [19]. Moreover, the meaning of these experiences is intrinsically linked to each individual's unique values and enshrined in the things that matter to them [11,13]. Morality is also social as far as these values are embedded in social practices [20]. Accordingly, the causes of morally problematic experiences can be manifold. It could indeed be the case that there is a questionable attitude from a health care professional causing this problematic experience, but it could also be a simple misunderstanding or miscommunication. This is why ethical inquiries interrogate moral experiences and ask questions about why we experience challenges. Moreover, embedded in this account of ethical inquiry is a distinction between morality as designating the experiential domain of human values and preferences and ethics, which designates a structured field of inquiry, open discussion, research, etc, about tensions and questions raised about human morality and moral experience itself [20,21].

Participatory action research is a promising approach to conducting ethical inquiries that respond to such moral challenges. This approach may be embedded in a diverse range of research methodologies, including qualitative research [22,23]. It differs from conventional research by calling for partnerships with stakeholders from local communities to understand the challenges they face and collaboratively identify ways to mitigate them [22,24]. These challenges may include moral challenges addressed in partnerships with patients. Participatory action research applied to an ethical inquiry is premised on the value of experiential knowledge and agency of patients and other stakeholders [22,25]. Stakeholders may be involved in defining a project's direction (ie, toward certain moral challenges), improving its methodology, interpreting results, identifying paths for action, and disseminating results [24]. In such projects, ethicists do not position themselves as authoritative experts, dictating how moral challenges should be understood and resolved [16]. Rather than giving their own moral opinions, ethicists provide opportunities for stakeholders to dialogue, engage in these steps of the project, and critically reflect on the values at stake [26]. Therefore, participatory action research in bioethics fosters democratic inquiry, social change, and stakeholder empowerment [22,26].

To the best of our knowledge, few participatory action research projects have been conducted on rare diseases and bioethics. One project, conducted in the United Kingdom, involved the co-design of a national program for genomic screening for rare diseases in newborns with health care professionals, researchers, ethicists, patient groups, and members of the public [27]. Genomic screening for rare diseases raises important ethical issues such as data use and governance, medicalization before illness symptoms, discrimination, and resource use [27]. Otherwise, no participatory project has directly reported an ethical inquiry in relation to the moral challenges experienced by patients and potential solutions that could alleviate these challenges. This gap thus provides an opportunity for

methodological innovation in ethical inquiry and an important avenue of investigation for rare disease research.

Objectives

This paper presents the process and outcomes of the participatory development of an ethics action plan for rare disease care. This action plan represents a toolbox that provides a review of multiple possible interventions for policy makers, hospital managers, practitioners, researchers, and patient associations to critically reflect on the key moral challenges experienced by patients with rare diseases and the ways they could be mitigated. Health care workers and patients are the primary beneficiaries of this action plan, which seeks to promote their ongoing collaboration in health care. This action plan reflects the ethical inquiry introduced above. It provides these actors with an intellectual space to reflect on how rare disease care can be improved, while calling for greater sensitivity to patients' values. Thus, it departs from traditionally authoritative governmental action plans, whose implementation leads to externally motivated changes in governance, service provision, and resource allocation.

This ethics action plan was developed with members of the rare disease community in Québec, Canada, in accordance with the importance of local contexts for participatory action research [22]. Two bioethics researchers, 3 patient partners, 2 clinical researchers, and a representative from the *Regroupement québécois des maladies orphelines* (RQMO, or Québec Coalition of Orphan Diseases) were involved in the development of the action plan as members of *the Ethics and Rare Diseases Working Group*. The action plan is also supported by scientific literature, gray literature from Québec, and semistructured interviews conducted with patients with rare diseases from this province. This action plan introduces an ethical inquiry into rare diseases that is informative, relevant, and potentially informative in other health care contexts [10]. The participatory process underlying the development of the action plan favors its relevance and usefulness in the lived experiences of patients with rare diseases [22].

This study first describes the participatory process that underlies the development of an action plan. The overall structure of the action plan was then presented. Subsequently, 4 components of the action plan are described: the key moral challenges encountered by patients (component A), a vision for patient partnership in rare disease care (component B), the courses of action prompted by this vision (component C), and promising interventions that could help address the moral challenges by enacting these courses of action (component D). These components, which emerge from qualitative methods, are presented along with commentaries that reflect the perspectives of the working group.

Methods

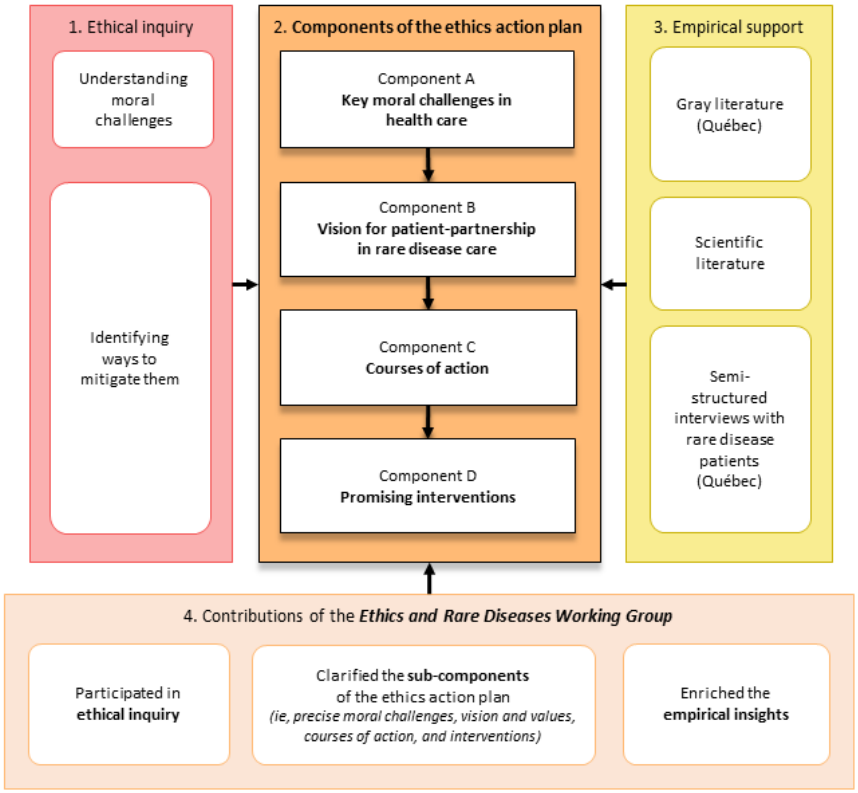
Overview

Figure 1 provides an overview of the development of the ethics action plan with the *Ethics and Rare Diseases Working Group* using qualitative methods. The resulting action plan was supported by insights from an exploratory literature review and a community survey conducted by our team [28] (Quintal A, unpublished data, September 2023). The structure of an action plan is inspired by the logic underlying ethical inquiry (Figure 1). It begins by identifying the key moral challenges. It then presents ways to address them through values tied to a vision for patient partnership in rare disease care, courses of action, and promising interventions (Figure 1).

The ethics action plan was developed iteratively by the first author, with significant input from the coauthors. The initial version of the action plan was derived from informally shared insights by working group members during previous meetings held by videoconferences dedicated to parallel qualitative studies [28]. The second and third versions of the action plan were drafted following consultations with the working group held via video conference and email. The action plan was subsequently improved based on the working group's feedback on manuscript drafts that progressively integrated insights from the exploratory literature review (eg, to further document certain types of interventions) and semistructured interviews (eg, to connect moral challenges and specific components to lived experience; Figure 1). Importantly, and consistent with the distinction drawn above between morality and ethics, a moral challenge is not a label for attribution of blame but rather a concept to designate something that puts at stake one's deeply held values.

An ethical inquiry approach is central to the development of an action plan. The first author, specializing in ethical deliberation methods inspired by pragmatist ethics [21], intervened as a mediator during consultations with the working group. When disagreements arose during the videoconference consultations, the first author invited each person to expand on their perspective and highlight the importance of the topic. On the basis of the shared information, she proposed a compromise that could be improved through additional discussion. If the disagreement persisted, she prioritized the perspectives of the patient partners, given the research team's commitment to emphasizing the lived experiences of individuals living with rare diseases. In contrast, no notable disagreements occurred during email consultations as the latter were mostly used to address minor details. Ultimately, the final action plan was approved by all authors via email. Both data collection methods were conducted by the first author with significant input from the coauthors.

Figure 1. Development of the ethics action plan through ethical inquiry, empirical support, and participatory involvement.



An exploratory literature review was conducted to provide empirical support for moral challenges, the vision for patient partnership in rare disease care, courses of action, and promising interventions in the initial versions of the action plan. This exploratory review covered the gray and scientific literature. Consistent with the local scope of participatory research, the gray literature review focused on documents addressing rare disease care, services, and policies in the province of Québec [6,29,30] (personal communication with the RQMO, 2023). Two unpublished documents were included in the gray literature review. The first document, developed in 2013, reports the full results of a provincial survey conducted by the RQMO. The second document, developed in 2017, presents a business plan to guide the expansion of rare disease clinics.

A scientific literature review was conducted through an exploratory approach using PubMed searches mirroring the components of the action plan. Additional scientific literature was consulted to clarify the topics raised in the previously identified literature. To this end, additional PubMed searches were performed, and references cited in the previously identified literature were consulted. All content relevant to the action plan was extracted in a dedicated word document and progressively synthesized in the form of the current article.

Semistructured interviews with adults living with rare diseases [31] were conducted from February to March 2022. The participants were recruited from among the respondents of a previous community survey conducted by our team. Briefly, this survey aimed to document the most morally problematic experiences encountered by individuals living with rare diseases in the province of Québec, Canada. The inclusion criteria for this survey included were being aged 18 years or older, living in Québec when the survey was conducted, and living with a diagnosed or an undiagnosed and suspected rare disease (Quintal A, unpublished data, September 2023). The respondents were recruited through convenience sampling [32]. The RQMO, along with more than 80 patient associations and support groups, was invited to promote the survey in their newsletters and social media pages. Following this effort, 246 questionnaires were initiated, and 121 were sufficiently filled to be included in subsequent analyses. A total of 95 rare diseases were identified among the respondents. Respondents' ages ranged between 18 and 79 years, and 78.5% (95/121) of the respondents were women (Quintal A, unpublished data, September 2023).

Interview participants were selected from among survey respondents to constitute a diverse sample with regard to age, rare disease, and region. Participants were invited to participate in the interview through an initial short email and were provided with more information, including a consent form, if they expressed curiosity or interest in the study. Among the 18 invited respondents, 12 agreed to participate in the interview. Interviews were conducted by phone or videoconference [33,34]. They were then recorded and subsequently transcribed using an external transcription service.

Simple thematic analyses were conducted on transcript excerpts secondary to the analyses made for another manuscript reporting the primary data (Quintal A, unpublished data, September 2023). Two coding guides were generated. In the first coding guide, which was generated by the first author, primary themes reflected the components of the action plan, which were key moral challenges encountered by rare disease patients (component A), the vision for patient partnership in rare disease care (component B), courses of action (component C), and

promising interventions (component D). Secondary themes corresponded to the subcomponents of the action plan [35]. This correspondence was approximately due to the evolving nature of the action plan. Relevant transcript excerpts were linked to secondary themes. The excerpts reported in this manuscript best illustrated these secondary themes. In the second coding guide, which was generated by a research assistant, primary themes and secondary themes were designated categories and subcategories of patient empowerment strategies, respectively. The second coding guide has been validated and enriched by the first author. Component C describes the empowerment strategies uncovered with the second coding guide.

Ethical Considerations

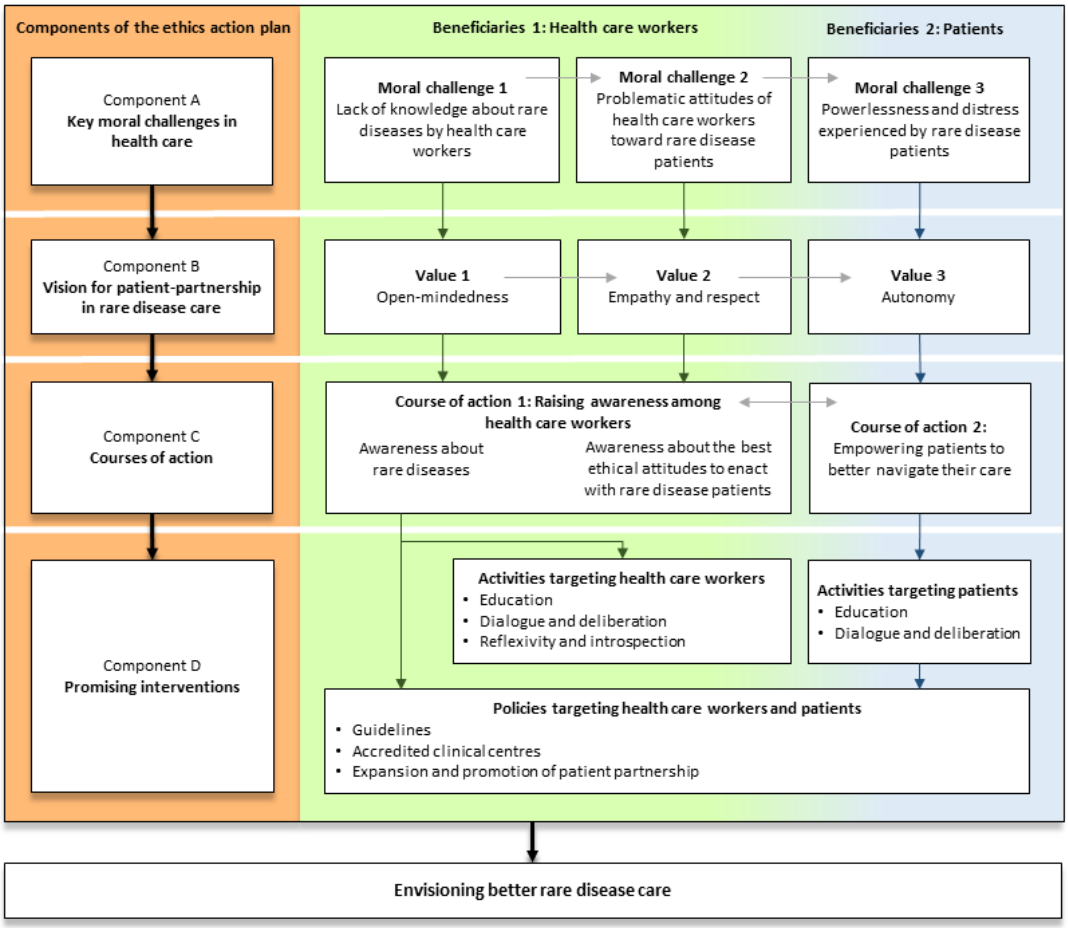
The study was approved by the Human Subject Ethics Committee of the Montreal Clinical Research Institute (2021-1080). It complied with the Standards on Research Ethics and Scientific Integrity of the *Fonds de recherche du Québec-Santé*, the Tri-Council Policy Statement 2—Ethical Conduct for Research Involving Humans of the Panel on Research Ethics of the Canadian government, and the Helsinki Declaration. The survey respondents provided informed consent before participating in the study. Interview participants were offered $50. Three $50 gift cards were drawn for survey participants. Survey and interview participants were deidentified (ie, by assigning a number to each participant). Transcripts were anonymized (ie, by removing all references to identifiable individuals, locations, etc).

Results

Overview of the Ethics Action Plan

As illustrated in Figure 2, the ethics action plan begins by highlighting the key moral challenges characterizing rare disease care (component A). The lack of knowledge about rare diseases among health care workers may contribute to problematic attitudes toward patients with rare diseases. Following these encounters, patients may feel powerless and distressed. These key moral challenges call for a vision for patient partnerships in rare disease care (component B). This vision is rooted in specific values: open-mindedness from health care workers, empathy and respect from health care workers, and autonomy for patients. Two courses of action target health care workers and patients to move toward these values: greater awareness about rare diseases and patients' empowerment in navigating their care, respectively (component C). Promising interventions, including activities and policies aimed at health care workers or patients, could foster these dispositions (component D). Ultimately, the ethics action plan aims to stimulate ethical reflection on improving rare disease care through greater sensitivity to patient values and concrete, constructive responses to their needs in health care practice changes.

Figure 2. An ethics action plan for rare disease care.



Component A: Key Moral Challenges in Health Care

Overview

Patients with rare diseases experience a plethora of moral challenges in health care. Three challenges stood out as most morally significant for members of the *Ethics and Rare Diseases Working Group* and for previously collected data [28] (Quintal A, unpublished data, September 2023). These challenges were the lack of knowledge on rare diseases among health care workers, health care workers' sometimes problematic attitudes toward patients, and the distress and powerlessness experienced by patients (Figure 2; component A). The following paragraphs further describe these challenges with relevant information from existing literature and supporting quotations from the interview participants.

Lack of Knowledge on Rare Diseases Among Health Care Workers

Unfamiliarity with rare diseases among health care workers is a key challenge in health care. Health care workers are poorly informed about the manifestations of rare diseases, the available services dedicated to rare diseases, and potential therapeutic avenues when available [29]. This gap in knowledge is unsurprising given that there are between 5000 and 8000 rare diseases worldwide, which have diverse causes and manifestations, including variability within the same rare disease [6,30]. Grasping the breadth and depth of knowledge about rare diseases is an impossible task for health care workers, who have already struggled with limited time and resources. However, health care workers have limited exposure to rare diseases in their training curricula or continuing education [6,29,30]. This could be due to the scattered and scarce scientific expertise on rare diseases [30].

This lack of familiarity with rare diseases among health care workers is a potential cause of the diagnostic odysseys experienced by patients. Misdiagnosis or the absence of a diagnosis may lead to improper medical treatment, unnecessary tests, or exposure to excessive radiation doses due to repeated scans. However, these inappropriate procedures can be expensive, useless, and dangerous [6]. Conversely, a formal diagnosis is usually required to access medications that may halt the progression of a rare disease or alleviate its symptoms. Without proper care, quality of life is compromised, sometimes leading to the death of patients [6,29,30]. Similarly, patients are denied disability benefits without a formal diagnosis [29], increasing financial struggles due to unemployment, and elevated costs of medication, care, and services, for example (Quintal A, unpublished data, September 2023). The multifaceted impacts of diagnostic odysseys elicit psychological distress for patients, adding to the distress they experience due to the lack of explanation for their health condition [6].

Health Care Workers' Sometimes Problematic Attitudes Toward Patients

Previous studies have illustrated that health care workers may display problematic attitudes toward patients with rare diseases. Patients report not being taken seriously [36-39] and express that health care workers do not believe in the severity of their symptoms [10,37,40]. They are sometimes labeled as hypochondriacs [41], or their symptoms are falsely attributed to psychological causes [36,40-42]. Patients are sometimes accused of lying [38,41] or are erroneously told that they invent their symptoms to receive attention or drug prescriptions [38]. Some patients report that health care workers lack compassion [43], belittle and stigmatize them [28] (Quintal A, unpublished data, September 2023), and do not listen to them [36]. Several factors may foster problematic attitudes among health care workers. These attitudes could stem from an unfamiliarity with rare diseases or personal dispositions.

First, health care workers may not be able to recognize their patients' disease manifestations. Overconfident health care workers may falsely assume that these diseases have psychological causes [44,45]. Alternatively, other health care workers may feel incompetent and resort to inappropriate defense mechanisms. They may project blame, frustration, and anger toward their patients [37,46,47]. Through this resentment, health care workers may also express close-mindedness by refusing to conduct further investigations or learn more about rare diseases [37,38]. By avoiding confidence in rare diseases, health care workers may further reinforce their problematic attitudes.

These factors were intertwined with the problematic attitudes of the participants. Blame, along with disbelief, was central to the testimony of a man aged 32 years living with empty nose syndrome. This condition is a rare and distressing complication of sinus surgery, resulting in loss of sensation of air passing through the sinuses. Upon carefully explaining his situation to numerous physicians, "they all said this: anxiety. They were all blaming it on my mental [state]." Later, he added that physicians told him, "You exaggerate. You are trying too hard. It's not that severe. You just want to bother everyone with this."

The 2 interview participants faced close-mindedness. A woman aged 46 years explained, as she sought a diagnosed with Ehlers-Danlos syndrome based on her severe symptoms:

> *I told [the internist]: "you know, I'm not here to tell you things or teach you stuff, but you likely know that there are many types of Ehlers-Danlos [syndromes]." She told me, 'Yes, there are six types.' I look at her, I tell her: "No, there are 14 types. There have been new criteria published in 2017. Would you like to see them?" [She said:] "No, no!" She pushed away my papers.*

Later, the participant added that the internist "did not want to learn about my story, my symptoms. She did not want to make a global [assessment of my condition]." A 29-year-old woman living with congenital hyperinsulinism shared a similar story: "among many physicians that I have met in my life, many are close-minded, you know, they are, like, convinced to have absolute knowledge, and they are not really curious to learn more."

Second, the problematic attitudes sometimes displayed by health care workers may stem from personal dispositions. Some health care workers may be naturally less empathetic than others, and contextual factors may lead them to express these attitudes while facing few negative repercussions. This reality is evidenced in

the story of a 47-year-old woman living with Sjögren syndrome secondary to rheumatoid polyarthritis in addition to a suspected disease of the pituitary gland who experienced aggression and violence from a physician [47]. To claim the disability benefits offered by her former employer's insurance company, she had to undergo evaluation by an expert physician hired as a consultant by the company. The physician was likely to have a conflict of interest: he had a disincentive to recognize her rare disease, as this would be costly for the insurance company that employs him as a consultant. These contextual constraints tainted his attitude toward her, as she explained:

> *He was aggressive. He manipulated the information. He wouldn't let me speak. I wasn't allowed to give information. I couldn't provide details. I had to answer by yes and no. I was subjected to a list of questions, I could not divert from it. He was aggressive, aggressive towards me. He yelled after me, he raised his voice. He basically kicked me out of his office by saying, "I'm done, you may leave." Yes, but me, I'm not done. I'm not done.*

Such conflicts of interest may arise in other chronic medical conditions or in long-term injuries. However, health care workers with little understanding of rare diseases may erroneously undermine the disability and incapacitation faced by patients and may not know how to properly assess patients with rare diseases. Health care workers risk harming patients if they handle these situations inappropriately, as the woman cited above recalled during her interview.

Distress and Powerlessness Experienced by Patients

Medical encounters riddled with tension, such as those described above, leave rare disease patients feeling deeply distressed, worried, and sad [28]. For example, the woman living with Sjögren syndrome described the residual emotional impacts of the consultation with the expert physician: "extremely hurt. Extremely broken. I am extremely disappointed with what I saw and experienced. Very disappointed. Very disappointed with the humanity of these people...It's a game that is too tough to play for me."

Through these medical encounters, patients feel profoundly powerless regarding the management of their rare diseases [37,41]. Several factors contribute to this limitation. For instance, patients might not be provided with optimal conditions for shared decision-making [10,38,43]. Often, they are not provided with suitable information regarding rare diseases. According to a survey conducted by the RQMO, 58.8% (144/245) of respondents received little to no information on their rare disease or the rare disease of their relatives (personal communication with the RQMO, 2023). Patients also experience powerlessness when at the mercy of inappropriate care and risk of death due to health care workers' unfamiliarity with rare diseases [41].

Equally worrisome, patients feel powerless regarding their illness management due to the internalized skepticism expressed by health care workers. Following the problematic attitudes of health care workers, patients may begin to doubt their illness experiences, symptoms, and rationality [37,40,48]. A woman living with Sjögren syndrome explained: "When you are faced with cruelty, you are screwed. Being yelled at, being ridiculed, and being laughed at in my face, it's like...it destroys you so much! This destroys you very much. You question yourself." This powerlessness may be enhanced by an uncertain prognosis, which often accompanies rare diseases [41]. Through the accumulation of these experiences, patients may ultimately lose trust in the ability of the medical establishment to provide appropriate care [10,37,41,49].

Component B: Vision for Patient Partnership for Rare Disease Care

Overview

In response to the challenges presented above, members of the *Ethics and Rare Diseases Working Group* proposed the vision of patient partnership in rare disease care. Patient partnerships bridge the medical and scientific knowledge of health care workers with the experiential knowledge of patients [25,50]. Patients acquire experiential knowledge through their first-hand experiences of their medical conditions, care trajectories, disability, occupational hardships, and associated psychosocial repercussions [25,51]. Many patients also acquire theoretical knowledge by consulting the medical literature on rare diseases or by attending conferences [52]. This study compensates for the limited information they receive from health care workers (personal communication with the RQMO, 2023).

Through patient partnerships in care, health care workers value patients' competencies and judgments [53]. Patient partnership in care contrasts with older models of care, where patients are merely provided with medical information and are involved by health care workers in care decisions [54,55]. In this approach, health care workers genuinely consider patients' perspectives and experiences rather than merely listening to them [52]. They support patients in making health-related decisions, managing their health autonomously, and acquiring health-related responsibilities in ways that are compatible with their needs and personal inclinations [50,52,53,56,57].

Working group members envisioned 3 guiding values for patient partnerships to respond to the 3 moral challenges previously identified. These values were open-mindedness from health care workers, empathy and respect from health care workers, and autonomy of patients (Figure 2; component B). These values could also work synergistically; positive attitudes of health care workers are likely to be conducive to the expression of patient autonomy. A 61-year-old woman living with chronic myelogenous leukemia explained: "communication and teamwork, partnership; in fact, this was a success. It will help the patient recover more quickly, leave the hospital more quickly, and have a better quality of life." In the next sections, these 3 values are described with supporting information from the literature and semistructured interviews.

Open-Mindedness of Health Care Workers

As a value, open-mindedness could help health care workers overcome their lack of knowledge of rare diseases. Patient partnership may only be achieved with this open-mindedness of the patients' experiential knowledge [49]. Through greater open-mindedness, health care workers may more readily suspect

rare diseases, educate themselves on rare diseases, and believe in patients' experiences.

With greater open-mindedness, health care workers would adopt a "pedagogy of doubt," a disposition promoted by French rare disease associations [30,58]. Health care workers can suspect rare diseases when faced with patients who exhibit atypical disease manifestations with no apparent cause. They could conduct further investigations into rare disease diagnoses rather than quickly dismissing patient complaints. They could refer these patients to specialists who can readily diagnose them under appropriate conditions [30]. The 32-year-old man cited above living with empty nose syndrome explained: "take the time to reject all [disease] possibilities. If you have not rejected them and you send [the patient] to the psychiatrist, the physical [ailment] is still here, but [they] are not in the right place. [They] are not at the right specialist." In addition to allowing for proper care, such investigations are important given the epistemic value of diagnoses for patients [59]. Diagnoses validate patients' experiences [60] while helping them make sense of their confusing and distressing symptoms [37].

With greater open-mindedness, health care workers are also more receptive to listening to patients. From patients' testimonies, health care workers can learn about rare diseases and their psychosocial impact. A 40-year-old woman living with hereditary angioedema echoing the suggestions of 2 other participants explained the following:

> *Listening is the most important factor...Rare diseases...are so rare that it is possible that you never encounter them in your life, so it is normal that [healthcare workers] do not know them. However, listen to the patient, at least! First, the patient who specifically has a rare disease, their disease, they will know it from A to Z. [The patient] will be able to explain the entire inflammatory process if needed...I am able to explain entirely what happens in my body. Listen!*

Open-minded health care workers are also more likely to believe patients' narratives. This was the main recommendation put forward by a 47-year-old woman living with Nutcracker syndrome in addition to other rare diseases: "I would simply like them to start by believing us."

This does not necessarily imply that health care workers should uncritically believe patients. Rather, health care workers should be attentive to the hardships expressed by the patient with a humane and humble demeanor. To their best ability, health care workers should value patients' perspectives and experiential knowledge. Health care workers should avoid disbelieving, stigmatizing, or belittling patients. Exchanges between health care workers and patients should be constructive and bidirectional, meaning that patients should approach the former with a positive attitude. Patient knowledge, which may align with medical knowledge, may help health care workers compensate for their lack of knowledge.

Often, health care workers' tendency to identify a given condition is influenced culturally. For example, despite its overwhelming rarity [61], psychosomatization is often erroneously suspected in patients [30]. This cultural bias is reflected by a recent issue dedicated to "disorders of somatic symptomatology" in *Le Médecin du Québec* in 2021, a medical magazine published by the *Federation of General Practitioners of Québec* [62]. Open-mindedness could guide health care workers to unpack biases that inform their practice. A 55-year-old woman with mast cell activation disorder, a lupus-like syndrome, and severe postviral syndrome resulting from COVID-19 emphasized the need for a culture change among health care workers. She critiqued their appeal to unfounded judgments and beliefs when evaluating patients rather than a rational and evidence-based approach.

Empathy and Respect of Health Care Workers

The value of greater empathy and respect could be fundamental for rare disease care [39]. Empathic health care workers are more readily able to imagine themselves in the situations of their patients and are concerned about their well-being [63-65]. During respectful encounters, health care workers value patients, give them adequate attention, and demonstrate esteem, courtesy, and consideration [66-68]. Without empathy and respect from health care workers, patients may not feel supported in making health decisions [49]. The importance of empathy and respect has been highlighted in the Québec government's recent Rare Disease Policy [6]. Empathy and respect can manifest as compassion, solidarity, kindness, or courage depending on the context.

For example, in our interviews, several participants expressed that they would appreciate more compassion from health care workers. A 31-year-old woman living with granulomatosis and polyangiitis explained that "we feel so vulnerable, I think that I would only need a little bit of human warmth, for real." She added that if health care workers are unable to express compassion at the patient's bedside, they could be accompanied by a nurse or social worker when announcing a life-changing diagnosis. Moreover, the 55-year-old woman cited previously emphasized the importance of cultivating courage among health care workers, which is essential to kindness and respect:

> *[We] have to explain to medical students that it's their duty to be here, even when it is difficult...It's part of their job to accompany human suffering. You can't want the patient to be somebody else's problem because it's a difficult case...So if [the patient] has the courage to be there, the healthcare professional should have the courage to be there as well.*

Autonomy of Patients

In alignment with this value, patients with rare disease can be supported in making health decisions autonomously [50]. Decisions made autonomously by patients reflect their personal preferences, converge toward their goals and aspirations, are contingent on the information at hand, and are grounded in their life trajectories [69]. Helping patients achieve a high level of autonomy in disease management is a key objective of patient partnerships in care [50].

Valuing autonomy does not equate to holding patients fully responsible or worse, blaming them [70,71]. Rather, health care workers could recognize that autonomy is contextual, meaning

that certain conditions may promote or inhibit a patient's capacity to act autonomously [69]. Health care workers may promote patient autonomy by creating a care setting conducive to these abilities [71,72]. For example, some conditions may be conducive to decisions that are voluntary (ie, free of coercion), deliberate (ie, supported by arguments open to critical examination), and supported by sound information [69]. A previously cited 29-year-old woman living with congenital hyperinsulinism lamented that the physicians who cared for her during her pregnancy were not familiar with her rare disease and gave her contradictory treatment advice. As a result, she could not fully enact her autonomy. She explained it as follows:

> *Well, first, I felt not being able to make an informed decision regarding [2 competing medications], since I had the impression that I was missing information and that the physicians provided contradictory information...As a result, you know, I did not have the opportunity to have a preference...for my decision.*

Component C: Courses of Action

Overview

The *Ethics and Rare Diseases Working Group* reflected on courses of action that would cultivate approaches that reflect the vision and values discussed previously in response to the key moral challenges identified. The first course of action would be to raise awareness among health care workers regarding rare diseases and appropriate ethical attitudes to adopt in rare disease care. A second course of action would be to empower patients to navigate their care better.

Raising Awareness Among Health Care Workers

In accordance with the first course of action, health care workers could be sensitized to the existence of rare diseases and appropriate ethical attitudes to adopt in rare disease care. This strategy could foster a context favorable to patient partnership in rare disease care by proactively defusing tensions inherent to clinical encounters in the context of rare diseases.

First, this course of action could address issues of misdiagnosis and disbelief by exposing health care workers to the idea that rare diseases truly exist and may be present among their patients. Awareness efforts could emphasize the diversity of rare diseases, their varied clinical presentations, and their often multisystemic repercussions. They could also showcase the usual life-altering consequences of living with a rare disease, ranging from psychosocial challenges to financial hardships [6]. Raising awareness could also reveal the ways in which these struggles are exacerbated by diagnostic odysseys and a lack of recognition from health care workers.

Efforts to raise awareness could also include providing health care workers with high-quality sources of information on rare diseases [6]. These sources include materials curated by scientific consortia (eg, the Orphanet portal) [6], research institutes (eg, the National Institutes of Health's Genetic and Rare Diseases Information Center) [73], and rare disease associations (eg, RQMO's iRARE Centre) [74]. By consulting these resources when faced with atypical patients, health care workers can progressively learn more about rare diseases.

These strategies to raise awareness could likely cultivate greater open-mindedness among health care workers and counter their lack of knowledge. Perhaps this could prove more effective than familiarizing health care workers with various training opportunities for rare disease characteristics they may quickly forget.

Second, health care workers can become acquainted with the life-changing impacts of their attitudes on patients. Beyond empathy and respect, these attitudes could also encompass compassion, solidarity, kindness, courage, and greater sensitivity to the peculiarities of rare disease patients' situations [6]. Health care workers could also be made aware of the distress and powerlessness experienced by patients, which often results from encounters where empathy and respect are lacking. When conveying these attitudes to health care workers, special attention should be paid to their expertise to avoid patronizing them.

Raising awareness about rare diseases and important approaches to rare disease care among health care workers also coincides with Axis 1 of the Québec government's Rare Disease Policy. This axis emphasizes the need to create or improve awareness of rare diseases among health care workers through training, knowledge translation, and improved access to information [6].

Empowering Patients to Better Navigate Their Care

The second course of action consists of empowering patients to better navigate the health care system and manage their health. Empowerment is the ability to take responsibility and exert control in decisions and actions regarding one's life, in the spirit of self-determination [71,75-78]. In health care settings, empowered patients seek information on rare diseases and health care systems. They make medical decisions by mobilizing appropriate resources and subsequently reflecting on the outcomes of these decisions [76,77].

Through their struggle for recognition, individuals living with rare diseases are known for their empowerment and resilience [79]. Paradoxically, quotations from the interview participants suggest that feelings of powerlessness coexist with empowerment strategies. It is plausible that the latter varies across individuals based on their personal dispositions and rare diseases. Thus, patients can plausibly benefit from the diverse empowerment strategies used by others. Consistent with the tenets of patient partnership in care, empowering patients can help them overcome their feelings of distress and powerlessness while reinforcing their autonomy [29,80]. Patient empowerment is also fostered by health care workers recognizing patients' expertise [81,82], supporting the expression of their preferences and autonomy [72,82], and providing them with the necessary resources to make health choices [71].

Following this course of action, patients could be exposed to empowerment strategies such as those uncovered in the survey study and interviews conducted by our research team. These strategies, which pertain to health care settings or personal health management, are summarized in Table 1 [28]. However, this does not constitute a comprehensive portrayal of all the empowerment strategies that may be used by patients. Patient empowerment aligns with Axis 3 of Québec's Rare Disease

Policy. Axis 3 highlights the importance of promoting research and innovation in the realm of rare diseases, notably through knowledge translation initiatives between researchers, health care workers, and patients, as exemplified in Table 1 [6].

Table 1. Patient empowerment strategies for health care settings.

Categories and subcategories of strategies[a]	Strategies evidenced in a previous survey study and interviews on rare diseases [28]
Patient capacities, states, and resources	
Improving health literacy	• Seeking information on rare diseases through web searches or with the help of rare disease associations
Skills and attitudes symbolic of autonomy in care	• Vigilance regarding treatments administered, especially during hospitalizations • Being resilient • Being combative about one's health • Being perseverant
Control over health and care	• Returning to the hospital to seek answers • Being accompanied by a close one or a representative of the hospital's users' committee
Patient behaviors	
Seeking medical care	• Turning to private health care • Contacting international experts or traveling to obtain treatment, sometimes with the help of a GoFundMe campaign • Consulting several health care workers despite long waiting times • Privileging consultations with open-minded health care workers • Being selective about the hospital to travel to • Seeking a second medical opinion
Taking an active role in health care consultations	• Bringing written personal information to health care workers (eg, medical history, symptoms, possible diagnoses, and recommended and contraindicated drugs), while avoiding sharing information that could fuel their prejudices • Bringing scientific documentation to health care workers • Writing a letter to a specialist • Discussing medical interventions with health care workers • Looking for other medical treatments
Health self-management	• Carefully managing one's rare disease and symptoms • Using various strategies to access medical devices • Developing a protocol for one's rare disease • Complying with a prescribed treatment despite doubts about its appropriateness
Making medical decisions	• Choosing to opt out from a prescribed treatment • Opting out of medical consultations due to dissatisfaction
Seeking reparation	• Lodging complaints

[a]Both are inspired by the framework on patient empowerment developed by Bravo et al [77] and the literature cited in this paper.

Component D: Promising Interventions

Overview

The previously proposed courses of action, which raise awareness among health care workers and foster patient empowerment, may be realized through various interventions. Interventions are classified as activities targeting health care workers or patients or as policies based on a generous interpretation of a typology coined by Michie et al [83,84]. A critical analysis of the most promising interventions based on ideas from the *Ethics and Rare Diseases Working Group* and insights from the literature is presented below. This action plan represents a toolbox that provides a review of multiple possible interventions for policy makers, hospital managers, practitioners, researchers, and patient associations to develop context-appropriate interventions for improving rare disease care.

Promising Activities for Raising Awareness Among Health Care Workers

Health care workers could be sensitized to the existence of rare diseases and educated on appropriate ethical attitudes to enact with patients through activities related to education, dialogue, deliberation, or reflective activities (Table 2). These activities can be organized by ethicists, social workers, social scientists, health care workers, and, sometimes, in partnership with patients.

Table 2. Promising activities to raise awareness among health care workers.

Type of activity and promising activities	Description	Limitations	Benefits
Educational activities (examples focus on continued education)			
Training activity with an evaluative component	Professors, patient representatives, or patients would assess the ethical attitudes of health care workers.	Knowledge and practices of primary care physicians may not improve following patient feedback [84-88].	Enrollment is incentivized by the scarcity of continuing education credits with an assessment component [89].
Talk by patients	Patients would share challenging or positive experiences while emphasizing the need for appropriate ethical attitudes.	Patients may feel uncomfortable sharing negative experiences.	Such talks may capitalize on existing activity with regular attendance such as clinical grand rounds [90].
Talk by a health care worker living with a rare disease	A health care worker living with a rare disease would sensitize health care workers to rare diseases and the need for appropriate ethical attitudes.	The health care worker may face professional prejudice by exposing a personal vulnerability.	Knowledge uptake may be more effective through a talk led by a health care worker as opposed to a patient. The former may bypass the prejudice of his or her peers.
Article followed by training session	A short article published in the newsletter of a professional medical association could raise awareness on rare diseases and advertise an upcoming webinar on appropriate ethical attitudes.	Health care workers may only skim through the newsletter, limiting its ability to raise awareness.	Professional medical associations have wide readership, favoring good exposure to such articles.
Dialogue and deliberation			
Collaborative learning	A group of health care workers, which possibly includes patients, would meet regularly [91]. At each meeting, an alternating health care worker would describe complex situation involving a rare disease patient. Others are invited to provide input on the case [92].	These activities require regular time commitments and are difficult to scale. These activities may not interest the health care workers which exhibit the most problematic attitudes.	Collaborative learning fosters critical reflection, mutual learning, and improved understanding, especially with patient involvement [91,93].
Pairing programs	Health care workers would be put in relation with rare disease patients and their families. They would exchange freely with them outside of health care settings during a few meetings [94].	Same as previous	Health care workers may become more confident and less anxious to interact with rare disease patients [63,95,96].
Reflective activities			
Moral case deliberation	A team of health care workers would reflect on the moral disagreement or uncertainty they face together in a situation [97,98] involving a rare disease patient. An ethicist would help them to recognize the values at play and identify solutions [97-99].	The ethicist only initiates this process if prompted by health care workers. Yet, the latter may not be fully sensitive to moral disagreement or uncertainty.	This activity fosters moral competency [97,98].
Debiasing activities	These activities aim to reduce biases [63,100,101]. Health care workers would reflect on how they would act in fictive situations involving rare disease patients. Then, they would be exposed to appropriate ethical attitudes [63], prompting them to critically reflect on their biases and attitudes [63,102].	Health care workers which hold the most biases are the least likely to enroll in the activity.	Debiasing may be very effective in fostering changes by eliciting strong emotional reactions (eg, shame) [62,101].
Autoethnography	A few health care workers would write personal narratives about difficult situations they faced with rare disease patients. They critically analyze their narratives through qualitative methods [103,104].	Autoethnography is particularly time- and resource-intensive. Insights may not be entirely transposable to future situations.	If presented during a clinical grand rounds session [104], the narratives benefit the audience in addition to those who have undertaken the autoethnography.

First, educational activities may be delivered through a variety of formats, namely, webinars, workshops, written materials, and sessions within clinical grand round schedules [90]. These activities could be offered at various training stages, such as during entry-level studies [29,30], residency [30] (personal communication with Yves Berthiaume, 2023), or as continuing education [30]. Activities offered as continuing education may more readily counter systematic misunderstandings or prejudice disseminated over time through the hidden medical curriculum [104-106]. In particular, continuing education has been shown

to improve knowledge among general practitioners [84,107]. Four promising continuing education activities are listed in Table 2.

Second, activities centered on dialogue and deliberation may provide spaces for health care workers to learn together. Alternatively, they may provide an opportunity to learn from patients without constraints in the clinical setting. Unlike educational activities, they require periodic involvement instead of being offered as a single session. Two promising dialogic and deliberative activities include collaborative learning and pairing programs (Table 2).

Third, during reflective activities, health care workers would "explore or examine a situation, an issue, or a particular object on the basis of their past experiences to develop new understandings that will ultimately influence their actions [and] challenge the practices, roles, beliefs, and values of practitioners" [108]. Difficult situations experienced by patients with rare diseases can be targeted through these reflective activities. Reflection is central to 3 promising activities: moral case deliberation, debiasing, and autoethnographies (Table 2).

Promising Activities for Fostering Patient Empowerment

Patients learned empowerment strategies through activities related to education, dialogue, and deliberation (Table 3). These activities could feature the empowerment strategies discussed in Table 1, or rare disease patients could share their personal strategies. In Québec, patient empowerment activities would benefit from being delivered in French to compensate for the lack of rare disease information offered on the internet in this language [30]. These activities could be designed or facilitated by patient experts, patient association representatives, ethicists, or social workers.

Table 3. Promising activities to foster patient empowerment.

Type of activity and promising activities	Description	Limitations	Benefits
Educational activities			
Short explanatory documents	These documents would introduce empowerment strategies with illustrations, diagrams, and positive language. They could be shared on social medial, in newsletters, or through pamphlets distributed in waiting rooms.	Efforts needed to diffuse these documents should not be underestimated.	They can reach many patients.
Short informational videos	These videos would use clear visuals and concise language to discuss empowerment strategies. They could be shared on social media and newsletters.	More resources are required to develop videos than documents.	Same as previous.
Detailed articles	Detailed articles addressing empowerment strategies would be published in patient-oriented magazines. An example is the *Magazine Expériences* in Québec [109], a magazine produced by patient partners.	These specialized magazines have a limited readership.	Such articles enable patients to share their empowerment strategies and conversely, allow readers to become familiar with them.
Workshops	Workshops, offered virtually or in-person, would expose patients to documented empowerment strategies. They could include a subsequent breakout session to allow patients to share their personal strategies.	No limitations identified.	Workshops integrating theoretical content and patient interactions are informative and dynamic; web-based workshops accommodate geographically dispersed patients or those living with physical limitations.
Dialogue and deliberation			
Regular empowerment meetings	Patients would discuss empowerment strategies during regular group meetings. They would update others on their use of these strategies in ongoing situations.	Such meetings require regular time and energy investment from patients.	This activity provides a safe space for discussion among patients. It builds capacity and cooperation.
Patient companionship	Undiagnosed rare disease patients are paired with diagnosed patients. Pairs would be prompted regularly to discuss alternating empowerment strategies.	Discussions may drift away from empowerment as they occur privately within the pairs; this mentorship dynamic may be demanding for the experienced patient.	This activity fosters close relationships centered on peer support.

First, educational activities can be implemented or advertised by rare disease associations, moderators of online support groups, or health care workers. These activities include short explanatory documents, short informational videos, detailed articles, and workshops (Table 3). Second, activities centered on dialogue and deliberation could be developed through partnerships between various professionals, such as social workers, occupational therapists, or ethicists, and patient associations or moderators of online support groups. Dialogue and deliberation are explicitly directed toward the topic of empowerment, thereby surpassing the scope of existing group and peer support initiatives. These activities could preferably

be offered virtually to accommodate the geographic dispersion or physical limitations of the patients. Regular empowerment meetings and patient companionship activities are examples of these activities (Table 3).

The patient empowerment activities described in Table 3 overwhelmingly emerged from the discussions with the *Ethics and Rare Diseases Working Group*. Most patient empowerment activities described in the literature aim to improve illness management and clinical outcomes [82,110,111] (eg, therapeutic educational programs in France [30]) rather than to empower patients more holistically in their daily lives.

Promising Policies and Public Actions for Raising Awareness Among Health Care Workers and Fostering Patient Empowerment

Novel policies and public actions should be implemented to raise awareness among health care workers and promote patient empowerment. Policy development requires more resources than activities that target health care workers and patients. Nonetheless, promising policies include the development of guidelines for rare disease care, creation of accredited clinics, and expansion and promotion of patient partnerships.

First, a guideline document addressing the commonalities between most rare diseases could present the common challenges encountered by patients, emphasize the often unusual clinical presentations of rare diseases, and introduce appropriate ethical attitudes to enact with these patients [6]. These guidelines could also direct health care workers toward high-quality sources of information on rare diseases (Figure 2; course of action 1). These guidelines could also include references to local patient associations and online support groups [39]. With these guidelines, health care workers would be better equipped to direct their patients toward appropriate resources, as they undergo more detailed clinical investigations.

Second, accredited clinical centers for rare diseases could be created to unite several clinicians, benefiting patient care and interprofessional collaboration. Clinical centers could receive accreditation provided that their health care workers receive proper training on rare diseases and on appropriate ethical attitudes, notably through the above-mentioned educational activities. Despite being resource-intensive, this accreditation strategy would ensure the uptake of appropriate ethical attitudes among health care workers employed in these clinics and favor high-quality care [30].

Third, patient partnerships should be expanded and promoted for policy making and research. Such approaches contrast slightly with the vision of patient partnerships in care. Patient partnership in policy making or research involves integrating patients into management teams or research groups [30,51,53] beyond health care settings. Patient partners provide valuable inputs on paths for improving health care and health services. They also contribute to drafting policies and guidelines by mobilizing experiential and theoretical knowledge [30,51,112]. Although patient partnerships in research and policy making are gaining traction in Québec [112,113], they warrant greater extension to rare disease patients. As patients are not always knowledgeable about patient partnership initiatives, simple interventions could be implemented to enhance their exposure to these patient partnership opportunities, notably through flyers made available in waiting rooms or web-based advertisements. Although patient partnerships may require additional resources, the expansion and promotion of patient partnerships could help counter the marginalization experienced by patients with rare diseases. Our open experience in doing this is telling in terms of the ability of such partnerships to carry our research much further.

Discussion

This paper presents a first ethical action plan for improving the quality of care offered to individuals living with rare diseases. The action plan is articulated around 4 components: the key moral challenges encountered by patients (component A), a vision for patient partnership in rare disease care (component B), courses of action prompted by this vision (component C), and promising interventions that could help address moral challenges by enacting these courses of action (component D). It is intended to serve as a resource and toolbox to help policy makers, hospital managers, practitioners, researchers, and patient associations critically reflect on the key moral challenges experienced by this patient population and on ways to address these challenges. This action plan was supported by insights from the *Ethics and Rare Diseases Working Group*, gray and scientific literature, and semistructured interviews. More broadly, the article reflects on the values underlying rare disease care and patient experience, in addition to health care workers' beliefs and behaviors.

Although this ethical action plan is anchored in sociocultural elements specific to Québec, the ethical inquiry it initiates may benefit other health care contexts and cultures. Hence, future research conducted in Québec or elsewhere could involve the design, implementation, and evaluation of the promising interventions identified in this study. These activities can be developed in collaboration with working groups comprising stakeholders and should be subjected to responsive evaluations. These evaluative processes, which are participatory in nature, aim to assess the value of clinical practices or programs and to identify potential improvements [114,115]. The political landscape in Québec, which is characterized by the recent publication of a governmental policy for rare diseases [6], will likely favor the dissemination of this ethics action plan and the development of such activities.

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Conflicts of Interest

None declared.

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Abbreviations

RQMO: Regroupement québécois des maladies orphelines (Québec Coalition of Orphan Diseases)

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Original Paper

Enhancing Exposure Treatment for Youths With Chronic Pain: Co-design and Qualitative Approach

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Abstract

Background: Increasing the access to and improving the impact of pain treatments is of utmost importance, especially among youths with chronic pain. The engagement of patients as research partners (in contrast to research participants) provides valuable expertise to collaboratively improve treatment delivery.

Objective: This study looked at a multidisciplinary exposure treatment for youths with chronic pain through the lens of patients and caregivers with the aim to explore and validate treatment change processes, prioritize and develop ideas for improvement, and identify particularly helpful treatment elements.

Methods: Qualitative exit interviews were conducted with patients and caregivers at their discharge from 2 clinical trials (ClinicalTrials.gov NCT01974791 and NCT03699007). Six independent co-design meetings were held with patients and caregivers as research partners to establish a consensus within and between groups. The results were validated in a wrap-up meeting.

Results: Patients and caregivers described that exposure treatment helped them better process pain-related emotions, feel empowered, and improve their relationship with each other. The research partners developed and agreed upon 12 ideas for improvement. Major recommendations include that pain exposure treatment should be disseminated more not only among patients and caregivers but also among primary care providers and the general public to facilitate an early referral for treatment. Exposure treatment should allow flexibility in terms of duration, frequency, and delivery mode. The research partners prioritized 13 helpful treatment elements. Most of the research partners agreed that future exposure treatments should continue to empower patients to choose meaningful exposure activities, break long-term goals into smaller steps, and discuss realistic expectations at discharge.

Conclusions: The results of this study have the potential to contribute to the refinement of pain treatments more broadly. At their core, they suggest that pain treatments should be disseminated more, flexible, and transparent.

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KEYWORDS

co-design; participatory design; pain; exposure treatment; youths with chronic pain; caregivers; qualitative analysis

Introduction

Background

Chronic pain is among the largest contributors to disability in children [1], and suboptimal responses to current treatments remain a challenge for researchers and practitioners [2,3]. Youths with chronic pain experience major barriers to accessing adequate pain treatment (eg, those owing to shortage of providers and geographical distance) [4,5]. When offered a multidisciplinary pain treatment, a substantial number of patients

decline to participate [6,7]. The reasons for this decline are largely unknown. Although there is evidence to support psychological interventions in pediatric chronic pain populations [3], there is room for improvement to enhance pain outcomes and emotional functioning.

Rooted in the fear-avoidance model [8], a graded exposure treatment (GET) was designed to more explicitly target maladaptive mechanisms of pain-related fear and avoidance to improve the return to function. GET has been shown to be an effective treatment for adults with chronic pain [9]. However, GET was associated with higher dropout rates than traditional cognitive behavioral therapy [9]. GET demonstrated preliminary efficacy in youths with chronic pain (GET Living) in an initial single-arm trial [7]. By 3- and 6-month follow-ups, >80% of participants showed improvements in the primary outcomes of fear and avoidance and secondary outcomes of pain catastrophizing, pain intensity, and pain acceptance. Analysis of data collected in a subsequent randomized controlled trial (RCT) to evaluate GET Living in a larger sample in comparison with a traditional multidisciplinary pain management approach is still ongoing [10]. Treatment delivery in the RCT shifted to a web-based format during the COVID-19 pandemic [11], with data collection concluding in January 2022 (Simons, LE, unpublished data, January 2022). Although this shift rapidly dispelled distance barriers, new issues related to adequate treatment delivery in patient homes emerged.

In the planning of the next GET Living iteration, we faced several questions that are also asked in the broader literature: How can pain exposure treatments be improved to produce long-lasting effects? How can we ensure that patients receive and participate in pain exposure treatments on a larger scale? Although the COVID-19 pandemic forced us to take unusual pathways, is the remote delivery format something we want to continue? Therefore, we decided to take an intermediate step to engage with people with lived experiences before deciding which action should be taken next.

Co-design is a "meaningful end-user engagement in research design that includes instances of engagement that occur at all stages of the research process and range in intensity from relatively passive to highly active and involved" [12]. Patients with lived experiences are engaged as consultants or partners in the research process (in contrast to traditional research participants) with the aim of collaboratively improving treatment efficacy, relevance, engagement, and delivery [13-15]. Participatory paradigms can be situated in implementation science, improvement science, and citizen science, although they often lack explanatory theories and models [15,16]. For example, outcome domains informed by expert guidelines do not necessarily represent meaningful domains for those receiving the intervention [17,18]. Similarly, clinicians and researchers risk limiting themselves in their understanding of treatment mechanisms depending on their preferred theoretical model [19]. It is possible that the mechanisms targeted and assessed during GET do not adequately capture all that might change for an individual during exposure treatment. Thereby, patients and caregivers with lived experiences can provide valuable feedback about how to improve treatment and what specific treatment elements were helpful in promoting change.

Goal of This Study

In this study, we partnered with patients and caregivers who had previously received GET Living treatment [20]. From an improvement science perspective [15], our aims were to (1) explore and validate treatment change processes, (2) prioritize and develop ideas for improvement (ie, to refine the GET Living program for in-person and remote delivery), and (3) identify particularly helpful treatment elements to promote change.

Methods

Overview and Design

Overview

This project comprises two parts: (1) semistructured exit interviews and (2) co-design meetings. Qualitative exit interviews were conducted with patients and caregivers during a discharge session after they received the GET Living intervention as research participants. Subsequent co-design meetings were held with the patients and caregivers as research partners to refine the intervention in a formative research process.

Setting

This project involves 2 separate examinations of GET Living: one was a single-arm trial (Boston trial) and the other was an RCT (Stanford trial). The Boston trial (NCT01974791) used a sequential replicated and randomized single-case experimental design (SCED) with multiple measures evaluate the effect of GET on youths with chronic pain for the first time [7]. The Stanford trial (NCT03699007) used a 2-group RCT enhanced with SCED elements to compare GET Living with a traditional multidisciplinary pain management approach [10]. The former GET Living participants had a unique expertise in what it is like to undergo pain exposure treatment from a patient's and caregiver's perspective.

Recruitment

In the Boston trial, patients were recruited from the Pain Treatment Service at Boston Children's Hospital between December 2013 and February 2017 (data collection was completed in January 2018). In the Stanford trial, patients were recruited from the Pediatric Pain Management Clinic at Stanford Children's Health from January 2019 to May 2021 (data collection was completed in January 2022). Treatment providers referred patients to GET Living during clinic visits. A study flyer and additional brochures were also available in the patient waiting room for patients to self-refer to the study. Patients were deemed eligible to participate in GET Living if they were aged 8 to 17 years, had a diagnosis of chronic pain, had moderate to high pain-related fear, and had moderate to high functional disability [7,10].

Part 1: Qualitative Exit Interviews

Goals and Overview

Interview data were analyzed to identify themes related to treatment change processes. In addition, interviews were conducted to create a pool of ideas for intervention improvement

and helpful treatment elements, which were later ranked and discussed in the co-design meetings.

Interviewed Patients and Caregivers

Only the patients and caregivers who completed all treatment sessions were included in the qualitative analysis to ensure that the data were reflective of the entire treatment experience. The interview that was conducted with a patient and their caregiver who withdrew their participation was excluded. Both the patient and caregiver felt that the treatment's focus on pain and anxiety was not a good fit. In the Boston trial, 26 interviews of patients and caregivers were analyzed. In the Stanford trial, 26 interviews of patients and caregivers who were randomized to the exposure intervention were analyzed. The patients and caregivers were interviewed separately. More details on the interviewed cohorts are presented in Table 1.

Table 1. Demographics and medical characteristics of youths who received GET Living[a] in the first (n=26) and second (n=26) clinical trial.

Variable	Boston cohort	Stanford cohort
Age (years)		
Values, mean (SD; range)	13 (3.12; 8-20)	14 (2.73; 8-18)
Sex, n (%)		
Female	20 (77)	24 (92)
Race, n (%)		
White	22 (85)	22 (85)
Black or African American	1 (4)	2 (8)
Multiracial	2 (8)	0 (0)
Asian	0 (0)	1 (4)
Unknown	1 (4)	1 (4)
Parent marital status, n (%)		
Married	21 (81)	20 (77)
Single	1 (4)	1 (4)
Divorced or separated	4 (15)	4 (15)
Widowed	0 (0)	1 (4)
Pain diagnosis, n (%)		
Musculoskeletal	9 (35)	21 (81)
Neuropathic	8 (31)	2 (8)
Abdominal	6 (23)	3 (12)
Headache	2 (8)	0 (0)
Headache and musculoskeletal	1 (4)	0 (0)
Duration of pain (months), n (%)		
Values, mean (SD; range)	22.6 (27.5; 1-65)	40.5 (37.1; 4-138)
FDI[b] at baseline, n (%)		
Values, mean (SD; range)	25.23 (10.3; 2-47)	23.15 (10.07; 4-42)
Fear of pain (FOPQ[c] total), n (%)		
Values, mean (SD; range)	50.96 (19.8; 9-82)	56.58 (15.9; 10-84)

[a]GET Living: graded exposure treatment for youths with chronic pain.

[b]FDI: Functional Disability Inventory.

[c]FOPQ: Fear of Pain Questionnaire.

Interview Guide

The semistructured exit interviews were conducted by research assistants during the discharge visit following the completion of GET Living. All research assistants were trained by the principal investigator LES. In the Boston trial, most interviews were conducted in person. In the Stanford trial, most interviews were conducted via phone or video calls. The patient and caregiver interview schedules both comprised 8 questions (Multimedia Appendix 1). The questions were intended to capture the positive (eg, question [Q] 1: "What did you like the best about GET Living treatment?" "What was the most helpful?") and negative (eg, Q2: "What did not help?" "What

would you change?") experiences that the families had during their treatment. The participants were also encouraged to give their critical feedback through several questions (eg, Q3: "What do you wish you had known before starting GET Living treatment?"). Other questions targeted to capture treatment change processes, that is, the changes that the families experienced in themselves (eg, Q4: "What did you learn about yourself and your family in GET Living treatment?"). The interview schedule questions guided the conversation; however, consistent with the semistructured nature of the interview, the participants were also provided with space to share additional feedback about their experiences.

Analysis of the Exit Interview Data

Reflexive thematic analysis [21] was used to assess the participants' perspectives and identify common themes across the interview data. Consistent with constructivist epistemology, reflexive analysis allows for the cocreation of knowledge between the participants and researchers. Subjectivity is not seen as a potential threat to the "*truthful*" or objective meaning of the data but is rather conceptualized as an analytical resource for data interpretation [22]. Data analysis was led by an investigator (LS) who was not involved in the data collection or intervention delivery. The analysis was conducted by following the 20-question guide by Braun and Clarke [22].

To begin data analysis, the investigator became familiar with the data by repeatedly and actively reading 12 fully transcribed interviews and listening to some randomly selected interviews. For the subsequent coding process, analysis was conducted on the audio recordings of interviews instead of the transcriptions to capture richer, more nuanced (eg, tone and affective aspects of responses) aspects of the participant responses. While listening, the investigator entered detailed notes of the codes for each interview into a comprehensive overview table. Relevant quotes were fully transcribed. Throughout the data analysis, the first author (LS) incorporated semantic features of the data (ie, explicitly stated ideas, concepts, meanings, and experiences) as well as latent features (ie, implicit meanings underlying explicit statements) when defining codes and themes. The generated codes were then clustered into candidate themes. This analytical process focused on the development of themes related to treatment change processes throughout the GET Living program. Theme identification occurred through an iterative process, whereby 2 authors (LS and LES) identified and refined codes and illustrative quotes until deep and nuanced themes regarding change processes were developed. Interview data regarding particularly helpful elements and ideas for improvement were organized into topic summaries (in comparison with fully developed themes). These topics summaries were used as a starting point to facilitate ranking and discussion in the subsequent co-design meetings. They will be presented when describing the results of the co-design meetings.

Part 2: Co-design Meetings

Goals and Overview

The purpose of the co-design meetings was to validate the developed themes related to treatment change processes (eg, regarding their meaningfulness) and reach a consensus regarding important ideas for intervention improvement and key treatment elements. Consensus was established in 6 independent co-design meetings (ie, the nominal group technique) held as 3 parallel meetings with patients and caregivers. This allowed us to establish consensus within groups (ie, consensus in 1 group) and between groups (ie, consensus in multiple groups) as an estimate of the representativeness of the opinions expressed. Patients and caregivers served as ad hoc consultants [15] and were compensated for their efforts (US $30 per hour). Their role was to validate the research findings of the previous thematic analysis and to provide feedback about the GET Living treatment from the receiver's end [15]. The procedures were preregistered in the Open Science Framework [23]. The GRIPP2 (Guidance for Reporting Involvement of Patients and the Public) checklist for patient and public participation in research guided quality reporting of the study results [24].

Patient and Caregiver Research Partners

Patients and caregivers who were randomized to the exposure treatment arm of the GET Living RCT (Stanford trial), including treatment completers and dropouts, were invited as research partners. Approximately one-third of the people invited accepted the invitation (10/33, 30% patients; 14/33, 42% caregivers). Research partners attended 1 of the 6 independent co-design meetings with parallel meetings for patients (meeting 1a: 4/10, 40%; meeting 2a: 3/10, 30%; meeting 3a: 3/10, 30%) and caregivers (meeting 1b: 4/14, 29%; meeting 2b: 5/14, 36%; meeting 3b: 5/14, 36%). All research partners were invited to a final wrap-up session (5/10, 50% patients and 8/14, 57% caregivers). More details on the research partners who attended the meetings are presented in Table 2.

Table 2. Demographics and pain characteristics of the patient (n=10) and caregiver (n=14) research partners who participated in the co-design meetings.

Variables	Youths with chronic pain	Caregivers
Age (years)		
Values, mean (SD; range)	17 (2.4; 10-17)	49 (5.3; 35-55)
Sex, n (%)		
Female	10 (100)	12 (86)
Race, n (%)		
White	9 (90)	12 (86)
Black or African American	0 (0)	1 (7)
Asian	1 (10)	1 (7)
Ethnicity, n (%)		
Hispanic	0 (0)	2 (14)
Not Hispanic or Latino	10 (100)	12 (86)
Unknown	0 (0)	0 (0)
Pain diagnosis, n (%)		
Musculoskeletal	8 (80)	N/A[a]
Neuropathic	1 (10)	N/A
Abdominal	1 (10)	N/A
Duration of pain (months)		
Values, mean (SD; range)	38.27 (17.3; 14-66)	N/A

[a]N/A: not applicable.

Procedure

The co-design meetings were scheduled for 2 subsequent calendar weeks (April 2022). The meetings were held via Zoom (Zoom Video Communications, Inc) and lasted approximately 120 minutes (including breaks). An optional web-based wrap-up meeting was held the following week (approximately 60 minutes). An overview of the procedure is provided in Multimedia Appendix 2. The meetings were moderated by CWH, LS, and LES.

Before the meeting, the research partners received a pre-engagement package with an outline of and the materials for the meeting. No preparation was required. At the beginning of the meeting, the research partners introduced themselves with ice-breaking tasks aimed at facilitating a good working atmosphere. Some ground rules were presented. Their role as research partners (as opposed to research participants) was highlighted.

The results of the thematic analysis of treatment change processes were then presented and discussed with the research partners to ensure that the identified themes were relevant and meaningful and to assess whether there were any important change processes missing from the established themes. The ideas for improvement collected during the semistructured exit interviews were then presented. The research partners were asked to rate the ideas using a Qualtrics (Qualtrics International Inc) survey. First, they were asked to select what they believed to be the 10 most important ideas out of the 48 ideas initially identified through the interviews. They were then asked to further refine their initial selection to identify the 3 most important ideas for improvement. The research partners were also encouraged to provide new ideas that were not found on the list as applicable. Once all the answers were collected, the results were shared with the group, and the research partners were asked to discuss the selections to ensure agreement among the retained items and address any differences of opinion regarding key recommendations. The same process was conducted to establish the most helpful out of 38 treatment elements that should be retained in future iterations of the intervention, and where applicable, the research partners were given intervention materials for review.

In the wrap-up meeting, action items from the co-design meetings were presented and finalized in a shared Word (Microsoft Corp) document. The research partners who could not attend the meeting were informed about the action items via email. They were asked to provide their written feedback within 2 weeks.

Evaluation of the Co-design Meetings

At the end of the meetings, all research partners completed module A of the Public and Patient Engagement Evaluation Tool [25]. Module A was developed to measure a 1-time engagement activity from a participant's perspective. The module consists of 13 statements (eg, "I had a clear understanding of the purpose of the co-design meeting"), which the research partners were instructed to rate on a 5-point Likert scale (1="strongly disagree," 2="disagree," 3="neither agree nor disagree," 4="agree," and 5="strongly agree"). The questionnaire also comprises 6 open-ended questions addressing

key elements of quality public and participant engagement, including the integrity of design and process, influence and impact, participatory culture and collaboration, and common purpose. The questionnaire was used as a quality measurement of research partner engagement in the co-design meetings. In addition, we openly asked the research partners why they agreed to participate in the co-design meetings.

Ethics Approval

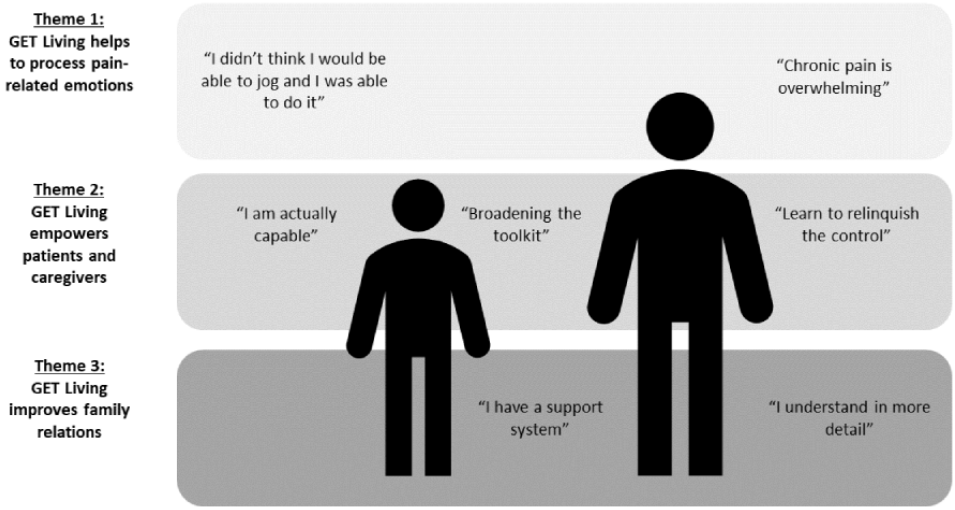
Both trials received ethics approval from their respective institutional review boards (Boston: IRB-P0000727, and Stanford University: Protocol 39514). Before their participation, the patients and caregivers actively consented to take part in the respective clinical trial. The final version of the manuscript was sent to the patient and caregiver research partners. All research partners provided their consent for publication.

Results

Part 1: Results of the Thematic Analysis

A total of 3 subordinate themes were generated from the reflexive thematic analysis of the exit interviews (Figure 1). These themes reflect the treatment change processes experienced by the patients and caregivers during the GET Living intervention. The themes were validated by the research partners in the co-design meetings.

Figure 1. Subordinate themes describing treatment change processes experienced by the patient and caregiver. The developed themes are summarized on the left. The subthemes are displayed within the boxes, with the subthemes derived for the patients presented on the left ("I didn't think I would be able to jog and I was able to do it" and "I am actually capable"), the subthemes derived for the caregivers presented on the right ("Chronic pain is overwhelming," "Learn to relinquish the control," and "I understand in more detail"), and the subthemes derived both patients and caregivers presented in the middle ("Broadening the toolkit" and "I have a support system"). GET: graded exposure treatment.



Theme 1: GET Living Helps Process Pain-Related Emotions

Overview

The first theme described how the patients and caregivers were better able to handle their pain-related emotions. Although the patients felt more confident in dealing with challenging situations, the caregivers had a space to process their own emotional struggles.

Patients: I Didn't Think I Would Be Able to Jog and I Was Able to Do It

The patients learned through exposures that the experience was not as bad or challenging as they thought it would be. Overall, the patients described an emotional shift in their experience because it did not match their expected outcome:

> *It was probably the first time they told me to go for a jog, I didn't think I would be able to do it, I got really scared but after I jogged with my mum it made me feel a lot better and I was able to do it and it made me happy.* [B9, patient]

Other patients and caregivers reported a change in their thinking:

> *Exposures were easier than I thought they would be.* [B30, patient]
>
> *The thought process going into it and getting those thoughts in check of how you can do things and not allowing misguided thinking to not allow you to do things that you can do.* [S24, caregiver]

There was a general shift in their perception of challenges. The patients also appeared to gain a sense of control:

> *I learned fear doesn't control me, I can control it and I can control how to deal with it.* [S15, patient]
>
> *I learned to not be so afraid of things I loved to do. There are some challenges but I can get through them.* [B20, patient]
>
> *Those exposures really assisted her achieving goals she didn't think she'd be able to make. And after that she was able to do more things.* [S24, caregiver]

Taken together, the patients realized that the situations they once feared were not as emotionally challenging or difficult as they expected them to be. On the basis of this experience, the patients seemed to see challenges as more approachable and manageable:

Caregivers: Chronic Pain Is Overwhelming

The caregivers became more aware of the overall experience of managing chronic pain and its impacts on the entire family:

> *Chronic pain is so overwhelming and such a challenge. Not only for the person in pain but the entire family really suffers from that...I don't think I recognized how bad it was until I got into the program.* [B19, caregiver]

> *I learned that my pain impacted everyone in my family not just me, so like, the pain, I might feel it but everyone else can experience it too.* [B29, patient]

The caregivers had room to express and process their own emotional struggles:

> *And some were just the sad or grieving things. Especially her age going into young adulthood.* [S46, caregiver]

> *And [clinicians] said, "Your daughter's gonna be okay. Her pain is real, but she's gonna be ok." That was really important for me to hear that. Finally, someone put it all together and made me feel like, okay she's not gonna break.* [GS33, caregiver]

Some even expressed a newly found admiration toward their children:

> *So, I guess I admire kids and people who get through the pain somehow, and it's without a break and they still manage. I guess my admiration for [child] and for people who've experienced that has increased.* [B33, caregiver]

> *And then, you know, I knew that [child] had it in him that he could push himself. I just think he needed help kinda pushing himself past that initial pain.* [S9, caregiver]

Taken together, the overwhelming experience of living with chronic pain was felt by the entire family. The caregivers mostly expressed feeling anxious or sad for their child; however, they were able to shift their perception by reinterpreting this struggle as a strength. Instead of feeling sad or anxious, they expressed an admiration for their children for handling painful situations.

Theme 2: GET Living Empowers Patients and Caregivers

Overview

The second theme described how the patients and caregivers felt empowered during treatment. Whereas the patients experienced becoming more confident, the caregivers gave their children more space to handle difficult situations by themselves. Both felt that they learned concrete strategies for navigating difficult situations.

Patients: I Am Actually Capable

The patients' experiences changed their perceptions of themselves. Many patients felt empowered and more confident. Some patients learned that they were capable of doing things despite their pain:

> *The only thing stopping me was myself...Well of course it was my back pain and all that. But I kind of held on to my back pain a little too much, for a little bit too long.* [S33, patient]

> *I learned that because I've been in a lot of pain and I put things off, that I am actually capable of doing a lot more.* [B25, patient]

> *I also learned that I can do anything even with my neck pain.* [S24, patient]

Other patients expressed a more generalized sense of being capable:

> *I am stronger than I thought I was.* [B31, patient]

> *I got more confident and stronger doing all the activities.* [B22, patient]

> *I learned that I was more determined and stronger than I thought.* [S17, patient]

Taken together, the patients changed their self-perception and appeared to be more confident in their ability to handle difficult situations in the face of pain and other challenges.

Caregivers: Learn to Relinquish the Control

The caregivers came to understand how to balance control and letting go. Some caregivers expressed that they could better see the benefits of giving their children more opportunities to handle their pain by themselves:

> *As parents, we do want to help out and control as much as we can, and to some extent I do still believe that we should be looking out for each other, you know, trying to prevent them from having pain, if it's possible. But if it's, in this kind of situation with the chronic pain thing, you learn to relinquish the control more and give them more options to handle it themselves.* [S3, caregiver]

> *I learned that [child] can be a lot more independent than I sometimes give him credit for. So sometimes I have to ease off in helping him. So, I think I learned that it is okay to let him tumble through something because then he will feel like he really did it himself.* [S60, caregiver]

Other caregivers reported that they became more aware of the negative effects of being overly controlling:

> *Well obviously, that we were holding her back from trying new things and not presenting things that would challenge or take her outside of her comfort zone. And we didn't realize what we were doing.* [B19, caregiver]

> *I learned that I can be pretty intense and anxious which contributes to my child's troubles or doesn't help her cope. I learned to be more relaxed. Was too rigid before, let go of that now.* [B5, caregiver]

Generally, the caregivers were able to hold back on responding with their initial reaction to better respond to the specific needs of their children:

> *For me it was really about my own responses to her and how to control my responses and be more*

> *understanding of where she was at. I was so caught up in my own being tired and stuff that I was going through. I really had the opportunity to stop and take a look and understand how much she was impacted and how I could really help her.* [B19, caregiver]

Taken together, the caregivers were better able to control their initial reactions. After reflecting on the consequences of their own behavior, many caregivers said that they needed to relinquish some control to empower their children to manage challenging situations independently.

Patients and Caregivers: Broadening the Toolkit

The patients and caregivers broadened their knowledge of concrete strategies to better cope and live with chronic pain:

> *But also offering additional strategies for feeling like we don't have to be helpless in the face of the pain when it's severe. I think that was super helpful to both of us.* [S22, caregiver]

> *I liked the emphasis on creating a sense of confidence and broadening the toolkit for dealing with pain and the headaches. Kind of finding ways to carry forward. To make life worth living, kind of, you know, preserve some quality of life.* [S22, caregiver]

> *With strategies of knowing how to cope with pain, okay, I am gonna be able to approach those challenges of life more so because I have that tool under my belt that says, oh you are hurting today, you are not having a good day, how are you gonna get going and in the end come out the other side successful.* [B23, caregiver]

The patients learned to break down activities in pursuit of long-term goals:

> *I learned that taking a [small] step at a time can help me improve so much more than trying to take a big step.* [B36, patient]

> *I learned to just make accommodations instead of stopping the activity altogether.* [B30, patient]

> *I think her learning how to set goals that are achievable and measurable. And for her to be able to make them so that they are realistic. So, it was really her individualized goals. She made them up and she decided with the team where her values were.* [B19, caregiver]

The caregivers felt that they had solid action plans to encourage activities:

> *To give me a bit more vocabulary or instructions how to talk about things, like..."remember you wanted to do this because of the goals you set for yourself."* [S60, caregiver]

> *And maybe just validate that we get it and when she does something that we recognize that. It does really help because when you are in the middle of it, you don't really think about how we are going to react and it changes how she feels.* [B19, caregiver]

> *It empowered us as parents to say, we know your pain is real, we know it might cause a little bit of back pain, and you can take breaks. It gave us strategies for what we can say and what we can do to help encourage her still do her everyday activities.* [S33, caregiver]

Taken together, the patients and caregivers learned concrete strategies to navigate through difficult situations. These strategies helped reduce feelings of helplessness in both the patients and caregivers.

Theme 3: GET Living Improves Family Relations

Overview

The third theme described how GET Living helped improve the relationship between patients and their caregivers. Being able to better understand the complexity of chronic pain, the caregivers were more able to validate their child's experiences and felt closer to them. The patients also indicated that they felt more supported by their caregivers.

Caregivers: I Understand in More Detail

The caregivers better understood their child's pain experience in their day-to-day difficulties:

> *I knew that she was hurting every day and that lots of things were difficult for her, but I think that I understand in more detail that even simple tasks, how and why they are difficult for her.* [S57, caregiver]

> *[Clinicians] taught me a lot regarding just [child]'s pain and how it can really, I don't know, change her behavior. In that if [child] is grumpy or tired. I never associated the pain with her emotions before, neither did my husband. So, it was really eye opening for us to understand the correlation.* [S24, caregiver]

The caregivers also understood the driving mechanisms of pain chronicity in more detail:

> *I was also kind of surprised in the session when he was doing the soccer practice because he kind of attributed the time when all his leg pain started with soccer, even though soccer did not, you know, cause it.* [S60, caregiver]

> *It took away my anxiety that it will hurt, but it won't harm her. The program made her try something. And some of the things she did, I knew that she would hurt herself. Not harm herself, but hurt herself.* [B19, caregiver]

The caregivers also reported being better able to validate the experiences of their child:

> *It never dawned on me before about how [child] could be feeling about this because no one can see it. And we just gave her a hard time about school and that she is not feeling it. And sometimes with that they have to keep validating it and hold on to it. And maybe just validate that we get it and when she does something that we recognize that. It does really help because when you are in the middle of it, you don't*

> *really think about how we are going to react and it changes how she feels.* [B19, caregiver]
>
> *My mom and dad...actually knew how I feel now and what I was going through.* [B20, patient]

Taken together, the caregivers became more aware of the difficulties of their children. They better understood the impact of chronic pain on pain-related disability and distress. The caregivers also became aware of the driving mechanisms of chronic pain, including emotional responses and misattributions. This allowed them to validate their child's experience more.

Patients and Caregivers: I Have a Support System

The patients and caregivers reported that GET Living fostered improved family connections. The patients became more aware that they were not alone because they had their caregivers and families to support them:

> *And that my family can help me do whatever, that I don't just have to rely on myself to help these things. I have a support system.* [S33, patient]
>
> *I learned that my family are very enthusiastic and willing to do those things with me.* [S22, patient]
>
> *And my family, I think, learned if I am in pain how they can help me deal with it.* [S15, patient]

The caregivers also felt a closer connection with their children:

> *We kind of had a better connection than we did before. Not that we had a bad connection, it's just the drives to the sessions.* [S9, caregiver]
>
> *I felt like some of the sessions led to more discussions with [child] and I afterwards, like I felt that there were certain things, like as a mother daughter, that it was positive.* [S46, caregiver]

Taken together, the relationships between the patients and their families improved. While the patients felt supported, the parents felt a closer connection with their children.

Part 2: Results of the Co-design Meetings

Ideas for Improvement

A total of 12 ideas for improvement were prioritized in multiple groups (ie, between-group consensus) and are presented in Table 3. The ideas were organized based on the degree of consensus between the groups. Five ideas that were prioritized by within-group consensus are presented in Multimedia Appendix 3.

Interestingly, these improvement ideas were not specific to exposure treatments and could be applied to any form of behavioral or physical pain treatment. For example, the research partners agreed that pain exposure treatment should be disseminated more. There was absolute consensus (consensus in 6/6, 100% co-design meetings) that pain exposure treatment should use patient testimonials to (1) provide patients with narratives of how other patients are dealing with similar difficulties, (2) inform future patients about what treatment will be like, (3) provide a role model, and (4) promote positive expectations. Most research partners also agreed that more efforts should be made to create awareness among the general public and primary care providers to facilitate an early referral for treatment (consensus in 4/6, 67% co-design meetings).

Table 3. Ideas of improvement developed and agreed upon in the co-design meetings using the nominal group technique (consensus in multiple groups)[a].

An ideal GET Living[b] program would...	Concrete ideas	Consensus between groups (n=6), n (%)
...inform what the treatment will be like and promote positive expectations	• Patient and caregiver testimonials (eg, videos) to see other patients dealing with similar difficulties, provide a role model, better understand what the treatment will be like, transmit hope for future patients • Clarify that the treatment aims to increase activity and explain the role of PT[c] (compared with traditional PT)	6 (100)
...start earlier with more interdisciplinary exchange	• More awareness of the program through posters, flyers, websites, and social media • Campaign educating primary care providers about this modality as a treatment option to facilitate early referral • More exchange and referral between providers (eg, to discuss treatment progress)	4 (67)
...allow for more flexibility	• Adapt the duration, frequency, and content to the momentary pain level or energy of patients • Flexible web-based sessions when pain level is too high	4 (67)
...be also offered remotely with optional in-person meetings	• Optional in-person meetings to build trust and help patients get a better diagnostic view of the exposure activities • Help overcome technical barriers (eg, send treatment materials at home and provide Wi-Fi booster)	3 (50)
...add booster sessions	• Combination of structured and client-lead booster sessions (eg, reminder of the core treatment elements and think together how they can be applied to real life)	3 (50)
...be honest that becoming better is not easy but it is a process	• Emphasize that treatment provides long-term strategies • Provide feedback on progress (especially little steps) as a motivator • Help to find the balance of being challenged but not overwhelmed	3 (50)
...have the patient decide if parent should participate in treatment	• Discuss with patients whether caregivers should join the treatment • Optional patient-only sessions	3 (50)
...be offered also to patients over 18	• Support in an especially vulnerable phase of transition into young adulthood (eg, decision on the future) on top of chronic pain	2 (33)
...enable patients to meet other patients	• Platform to exchange information with other patients of similar age (eg, ages of 8 to 12 years and ages 13 to 17 years) • Open coffee hours via Zoom (eg, once per month) • Web-based education sessions or booster sessions together with other patients	2 (33)
...enable parents to meet other parents	• Platform for support and exchange (eg, see other families who go through the same thing and think together how to positively influence family dynamics)	2 (33)
...include more complex pain ratings	• Description of end points (eg, developing individualized reference points at the beginning of treatment) • Body map to describe pain localization and give differential pain ratings for different locations	2 (33)
...be adapted to other symptoms experienced besides pain	• For example, adapt exposure activities to additional symptoms of dizziness • Editable worksheets to personalize exposure activities	2 (33)

[a]Ideas for improvement are organized according to the degree of consensus between groups.

[b]GET Living: graded exposure treatment for youth with chronic pain.

[c]PT: physical therapy.

Helpful Treatment Elements to Promote Change

A total of 13 treatment elements were considered helpful in promoting change in multiple groups (between-group consensus; Table 4). For a clear overview, helpful treatment elements are organized by treatment phase. Seven treatment elements that were considered helpful by only the members of 1 group are presented in Multimedia Appendix 4 (within-group consensus).

In general, the research partners appreciated the understanding attitude of clinicians, personalization of treatment through the

pursuit of individualized goals, education about chronic pain, encouragement of activities, and discussion of realistic expectations at discharge. For example, during the phase of goal setting, a majority of the research partners agreed that future exposure treatments should continue to empower patients to be "in charge" to choose meaningful exposure activities (consensus in 5/6, 83% co-design meetings), break long-term goals into smaller steps (consensus in 5 of the 6 co-design meetings, 83%), and help patients become aware of their own values and motivators (consensus in 2/6, 33% co-design meetings).

Table 4. Most helpful treatment elements agreed upon in co-design meetings using the nominal group technique (consensus in multiple groups)[a].

Treatment phase and future GET Living[b] programs (regardless of the delivery format) should continue to...	Consensus between groups (n=6), n (%)
Building rapport	
...combine pain psychology and physical therapy	3 (50)
...transmit the feeling that it is possible to deal with pain	2 (33)
...offer validation and understanding of patients' situation	2 (33)
Goal setting	
...empower patients to be "in charge" to choose meaningful activities	5 (83)
...distinguish between short-term and long-term goals	5 (83)
...help patients become aware of their values and motivators	2 (33)
Education	
...reflect on triggers of pain and anxiety	3 (50)
...distinguish between short-term and long-term solutions	2 (33)
...include the exposure graphs	2 (33)
Exposures	
...encourage activities allowing for breaks and a slow pace	3 (50)
...teach the use of facilitators	3 (50)
...include the WILD[c] scale	2 (33)
Discharge	
...discuss realistic expectations at discharge (eg, discuss coping with pain flare-ups)	6 (100)

[a]Treatment elements that were considered helpful are organized by treatment phases.

[b]GET Living: graded exposure treatment for youth with chronic pain.

[c]The WILD scale assesses a patients' perceived Willingness, Importance, Likelihood of Success, and Difficulty with regard to the chosen exposure. The scale is completed before and after exposure [10]. The WILD scale of an example patient can be found in Multimedia Appendix 5.

Evaluation of the Co-design Meetings

Overall, the co-design meetings were evaluated as good, with mean values being consistently at the upper end of the agreement scale (Tables 5 and 6). The research partners felt that the co-design meeting was a good use of their time, that they were able to contribute, and that they were confident that the meeting's goals were achieved.

Table 5. Quantitative results of the Public and Patient Engagement Evaluation Tool[a].

Item	Patients, mean (SD; range)	Caregivers, mean (SD; range)
Communications and supports for participation		
I had a clear understanding of the purpose of the co-design meeting.	4.3 (0.95; 2-5)	4.4 (0.63; 3-5)
The supports I needed to participate were available (eg, travel, childcare, etc).	4.5 (0.71; 3-5)	4.4 (0.76; 3-5)
I had enough information to contribute to the topic being discussed.	4.6 (0.52; 4-5)	4.6 (0.51; 4-5)
Views and perspectives		
I was able to express my views freely.	5 (0; 0-5)	4.9 (0.36; 4-5)
I feel that my views were heard.	4.9 (0.32; 4-5)	4.9 (0.36; 4-5)
A wide range of views on the topics discussed was shared.	4.5 (0.53; 4-5)	4.7 (0.47; 4-5)
The individuals participating in this co-design meeting represented a broad range of perspectives on the topic.	4.4 (0.7; 3-5)	4.5 (0.52; 4-5)
Impacts and influence of engagement initiative		
I think that the co-design meeting achieved its objectives.	4.7 (0.48; 4-5)	4.5 (0.52; 4-5)
I am confident the input provided through this initiative will be used by Biobehavioral Pediatric Pain Lab.	4.5 (0.71; 3-5)	4.6 (0.65; 3-4)
I think the input provided through this activity will make a difference to the work of the Biobehavioral Pediatric Pain Lab.	4.6 (0.7; 3-5)	4.6 (0.5; 4-5)
Final thoughts		
As a result of my participation in the co-design meeting, I am better informed about the Biobehavioral Pediatric Pain Lab.	4.4 (0.7; 3-5)	4.1 (0.77; 3-5)
Overall, I was satisfied with this engagement initiative.	4.6 (0.52; 4-5)	4.7 (0.47; 4-5)
This engagement initiative was a good use of my time.	4.7 (0.48; 4-5)	4.6 (0.65; 3-5)

[a]1=strongly disagree, 2=disagree, 3=neither disagree or agree, 4=agree, and 5=strongly agree.

Table 6. Qualitative results of the Public and Patient Engagement Evaluation Tool plus reasons for participation.

Open-ended questions	Patients	Caregivers
What else would you like us to know about how your participation in the co-design meeting was supported?	• Felt supported	• Easy web-based format with handouts given before • Accommodating and flexible scheduling
What else would you like us to know about how you were able to share your views?	• Everyone was easy to talk to • Everyone brought different perspectives and life experiences, which shaped their advice and made the discussion interesting	• Easier to share openly via Zoom
What else would you like us to know about the influence you think the co-design meeting will have?	N/A[a]	• Input may guide future improvements of an already great program
What were the strengths of the co-design meeting?	• Everyone was nice and supportive • Ability to contribute perspective on what to improve upon • Engaging and friendly leaders • Materials provided in advance • Surveys helped facilitate discussion	• Leaders were open and understanding, and our opinions were validated • Valuable to hear other perspectives from other patients • Able to voice concerns and connect with and hear the opinions of other caregivers • Smaller groups allowed for everyone's voice to be heard • Breakout rooms so that youths and caregivers could discuss separately • Informal nature allowed for comfortability
What could be improved about the co-design meeting?	• More icebreakers and introductions to meet the others in the meeting • Allow the patients to talk freely about their experience without structure to allow for suggestions that the researchers had not proposed and to allow the patients to connect with one another	• Would have liked a time to share freely without any structure
What else would you like us to know about your experience with the co-design meeting?	• It was a great way to allow the past patients to feel more included and important	• Allowed caregivers to hear others' experiences and thoughts
Why did you agree to be part of the co-design meetings?	• Wanted voice to be heard • Wanted to give back to a program that helped me • Wanted to help improve the program for others with chronic pain	• To help others with chronic pain • To give back and help this program • This study was very important to our family • Wanted to share my ideas for improvements

[a]No one answered the question.

Discussion

Principal Findings

Overview

This study looked at a multidisciplinary exposure treatment for youths with chronic pain through the lens of patients and caregivers. First, qualitative analysis of exit interviews conducted with patients and caregivers after they received the GET Living intervention explored the treatment change processes. Second, co-design meetings with patients and caregivers as research partners aimed to refine the GET Living intervention. The implications of both aspects are discussed in the subsequent sections.

Treatment Change Processes: What Changes and How?

The qualitative analysis revealed a wide range of treatment change processes, indicating that what happens within patients during treatment is complex and difficult to describe from a single theoretical lens [19]. The patients and caregivers described that the exposure treatment helped them to (1) better process pain-related emotions, (2) feel empowered, and (3) improve their relationship with each other. The elements of these reported changes align with different theoretical models. In line with the inhibitory learning approach [26,27], the patients experienced a violation of their expectations, wherein feared situations were not as emotionally challenging or difficult as expected they them to be. By contrast, the caregivers reported a reduction in protective behavioral responses when they felt more in control of their emotional distress, which, in turn, empowered the patients to handle difficult situations themselves. This aligns with the theoretical assertions of the interpersonal

fear–avoidance model [28,29]. In addition, the patients reported changes that are considered resources according to the resilience-risk model [30]. On an individual level, the patients reported improved self-esteem ("I am actually capable"). In terms of their family and social environment, they felt more supported ("I have a support system"). Consistent with the interpersonal process model of intimacy, the patients and caregivers experienced an increase in intimacy and improvement in their relationship when the caregivers were better able to understand and validate the patients' pain experience [31]. Looking through the lens of self-determination theory [32], the treatment might have satisfied the need for autonomy and competence (eg, by having patients be "in charge," which was ranked as a particularly helpful treatment element), which facilitated goal pursuit despite chronic pain and an increased a sense of confidence. The patients also felt more supported, indicating satisfaction in the need for relatedness. However, the patients wished to extend this support to their peers with chronic pain. Altogether, the present results underscore the need for a more holistic approach to understand the full complexity of treatment change processes within patients and in their interaction with their social environment. Future research using and combining contemporary quantitative methods (eg, ambulatory assessments, network analyses, and SCEDs) could use the present findings to flexibly and rigorously study treatment change processes from idiographic and nomothetic perspectives [33].

Refinement of Pediatric Pain Treatments: What Should We Do Better?

The research partners prioritized 13 core treatment elements that were helpful in promoting change. This feedback can directly inform clinicians which specific behaviors and techniques are perceived as impactful. This is informative for clinicians in general but especially in settings with time constraints. For example, a majority of the research partners agreed that future exposure treatments should continue to assist patients in finding and pursuing meaningful goals. This recommendation agreed with pain scientists, who advise combining exposure treatment with clarification interventions to identify personal goals and goal conflicts [34]. Such techniques could also have the potential to ameliorate other behavioral programs. However, future research should systematically investigate the benefits of these techniques (eg, improving outcomes or facilitating the transfer of skills to daily life).

The research partners also agreed upon 12 ideas for improvement. At their core, these ideas suggest that pain treatments should be disseminated more, flexible, and transparent. The research partners advised that there should be a platform for exchange between people with lived experiences and that the complexity of the individual pain experience should be acknowledged. To our surprise, most ideas were not specific to the content and refinement of exposure treatment; instead, they could inform the implementation of behavioral or physical pain treatments more broadly.

The research partners conveyed that increasing the access to and dissemination of pain psychology treatment is of upmost importance, a message also building momentum among pain scientists [35]. At the receiver's end, the research partners recommended to better clarify the role of psychological interventions in the context of a multidisciplinary pain treatment approach. Thereby, they came up with creative ideas such as video testimonials or advertising campaigns to clarify treatment aims and promote positive expectations. At the same time, the research partners also suggested better acquainting other treatment providers with this treatment option to facilitate early referral. In addition, they considered an increase in flexibility (eg, in terms of session duration, frequency, content, and delivery format depending on momentary pain level) a promising step toward improvement. Although shifting plans based on pain levels stands in contrast to pain scientists advocating that time and quota–contingent treatment plans are preferred over pain-contingent plans [36], it introduces an important consideration for pragmatic implementation in real life. Momentarily scaling back an activity versus rigid adherence to a plan could ultimately provide the flexibility needed to reach the long-term goal of greater life engagement and functionality.

The research partners considered the remote delivery format with optional in-person check-ins (eg, to build trust) as promising beyond the pandemic, which aligns with initiatives underway in the pain treatment field, as the pandemic has accelerated the dissemination of remotely delivered pain management services [37]. Continuing this path might contribute to a greater dissemination of pain psychology treatments, especially among youths. Remotely delivered treatments might also be beneficial for other behavioral or physical treatments (eg, to facilitate integration into daily life). Moreover, the research partners wanted more support in transferring and maintaining learned strategies (eg, via booster sessions). This request suggests potential ways to address the issue that the effects of pain psychology treatments are often not stable over time [3]. Altogether, the research partners created an abundant set of ideas focused on improving the delivery of pain treatments. From a human-centered design perspective, the present results specify the needs of patients and caregivers [38]. Future research could use these ideas to investigate whether tailoring implementation strategies to end users' needs relates to better behavioral (eg, penetration) and perceptual (eg, acceptability) implementation outcomes [39]. For example, it would be interesting to see whether tailoring implementation strategies for pain treatments results in fewer people declining to participate and fewer dropouts.

Strength and Limitations

We provided an in-depth analysis of a specialized multidisciplinary exposure treatment for youths with chronic pain. Although we provided an overview of the change processes experienced by patients and caregivers, we could establish whether they contributed to the overall improvement (eg, increase in physical activity and school performance) using the methods we adopted. The patients and caregivers did not report having experienced treatment side effects, although it should be noted that we included only treatment completers in the thematic analysis. We also did not explicitly ask about treatment side effects. Our findings may not be generalizable to other behavioral pain treatments or pain populations, although it is

likely that the fundamental processes identified are cross-cutting. Multiple co-design meetings allowed us to establish consensus within and between groups. This can be taken as an estimate of the representativeness of the expressed opinions. However, the included research partners were not representative, even of the US population, in terms of underrepresented groups, with most research partners being White and female. The study was conducted within the US health care system, and the results may not be generalizable to other health care systems and countries. In Germany, for example, the distances between patients' homes and outpatient pediatric care centers are smaller, and the acceptance of internet-delivered treatments is rather low [40]. We did not present differential consensus ratings for patient and caregiver research partners because they were largely congruent. Only the involvement of caregivers during treatment was a critical point, where although the patients wanted less involvement, their parents wanted more involvement. The compromise developed in the wrap-up meeting was to negotiate the amount of involvement at the beginning of treatment (also depending on the patient's age) and offer patient-only sessions.

The Future of GET Living

The GET Living team is poised to iterate and implement the advice learned from the patients and caregivers as research partners. Planned modifications span 3 key domains: publicity and education, treatment delivery, and supporting families after treatment completion. We intend to develop video testimonials that weave in the ingredients the patients and caregivers defined as essential, namely the opportunity to process pain-related emotions, feeling empowered, and improving their relationship with one another. In addition to patient and parent testimonials, we would like to roll out an advertising campaign that targets both patient families and providers regarding the role of psychology in pain treatment and in some instances, more specifically, the GET Living treatment approach. These 2 publicity and education initiatives will better elucidate treatment aims, address misconceptions, and cultivate positive expectations regarding treatment. In the realm of treatment delivery, we have demonstrated in our latest clinical trial the capability to deliver GET Living remotely [11], and a clinical trial to implement a digital exposure intervention is underway ([41]; NCT05079984). Finally, we aim to devise approaches that will lead to lasting positive effects. We envision integrating booster sessions up to 1 year after treatment completion, potentially a combination of in-person and remotely delivered sessions. Moreover, we can leverage our developing digital content to push resources to patient families over time and provide a library of tools accessible long after treatment completion. Altogether, these research partner–guided changes will undoubtedly improve engagement and outcomes among youths with chronic pain. For the future of GET Living, we plan to establish patients and caregivers as standing members of an advisory board to facilitate a closer collaboration with them during the next iteration of GET Living.

Conclusions

This study has revealed several powerful implications that should be considered in future treatments and studies. The exit interviews with the patients and caregivers demonstrated the full complexity of treatment change processes. The research partners agreed that pain exposure treatment should be disseminated more, flexible, and transparent. These implications would not have been revealed if only traditional outcome and facility measures had been used. The clear and meaningful outcomes of this study strongly support the involvement of patients and caregivers in pain treatment manual developments and pain study designs.

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Authors' Contributions

LS and LES developed the ideas for the manuscript. LS conducted the thematic analysis, supervised by LES. LS and LES designed and planned the co-design meetings together with CWH, ARVO, KAB, and LEH. LS, LES, CWH, and ARVO conducted the co-design meetings. LS drafted the first version of the manuscript with inputs from all the authors. CWH mainly revised aspects of the thematic analysis. KAB mainly revised aspects of the patient engagement process. LEH and JAG mainly provided input on clinical implications. LES supervised the project. All the authors read and approved the final manuscript.

Conflicts of Interest

None declared.

Multimedia Appendix 1
List of semistructured interview questions.
[PDF File (Adobe PDF File), 104 KB - jopm_v15i1e41292_app1.pdf]

Multimedia Appendix 2

Co-design meeting schedule.
[PDF File (Adobe PDF File), 97 KB - jopm_v15i1e41292_app2.pdf]

Multimedia Appendix 3
Ideas of improvement developed and agreed upon in co-design meetings using the nominal group technique (consensus in one group).
[PDF File (Adobe PDF File), 113 KB - jopm_v15i1e41292_app3.pdf]

Multimedia Appendix 4
Most helpful treatment elements agreed upon in co-design meetings using the nominal group technique (consensus in one group).
[PDF File (Adobe PDF File), 102 KB - jopm_v15i1e41292_app4.pdf]

Multimedia Appendix 5
The Willingness, Importance, Likelihood of Success, and Difficulty scale.
[PDF File (Adobe PDF File), 1118 KB - jopm_v15i1e41292_app5.pdf]

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Abbreviations

GET: graded exposure treatment
GRIPP2: Guidance for Reporting Involvement of Patients and the Public
RCT: randomized controlled trial
SCED: single-case experimental design
Q: question

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Original Paper

Co-design of the Transgender Health Information Resource: Web-Based Participatory Design

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Abstract

Background: There is an urgent and unmet need for accessible and credible health information within the transgender and gender-diverse (TGD) community. Currently, TGD individuals often seek and must find relevant resources by vetting social media posts. A resource that provides accessible and credible health-related resources and content via a mobile phone app may have a positive impact on and support the TGD population.

Objective: COVID-19 stay-at-home orders forced a shift in the methods used in participatory design. In this paper, we aimed to describe the web-based participatory methods used to develop the Transgender Health Information Resource. We also described and characterized the web-based engagement that occurred during a single session of the overall design process.

Methods: We planned and conducted web-based design sessions to replace the proposed in-person sessions. We used web-based collaborative tools, including Zoom (Zoom Video Communications), Mural (Mural), REDCap (Research Electronic Data Capture; Vanderbilt University), and Justinmind (Justinmind), to engage the participants in the design process. Zoom was used as an integrated platform for design activities. Mural was used to perform exercises, such as free listing, brainstorming, and grouping. REDCap allowed us to collect survey responses. Justinmind was used to create prototypes that were shared and discussed via Zoom. Recruitment was led by one of our community partners, One Colorado, who used private Facebook groups in which web-based flyers were dispersed. The design process took place in several workshops over a period of 10 months. We described and characterized engagement during a single design session by tracking the number of influential interactions among participants. We defined an influential interaction as communication, either verbal or web-based content manipulation, that advanced the design process.

Results: We presented data from a single design session that lasted 1 hour and 48 minutes and included 4 participants. During the session, there were 301 influential interactions, consisting of 79 verbal comments and 222 web-based content manipulations.

Conclusions: Web-based participatory design can elicit input and decisions from participants to develop a health information resource, such as a mobile app user interface. Overall, participants were highly engaged. This approach maintained the benefits and fidelity of traditional in-person design sessions, mitigated deficits, and exploited the previously unconsidered benefits of web-based methods, such as enhancing the ability to participate for those who live far from academic institutions. The web-based approach to participatory design was an efficient and feasible methodological design approach.

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KEYWORDS

transgender; gender diverse; participatory design; web-based design; co-design; health information resource; smartphone; app; mobile phone

Introduction

Transgender and Gender-Diverse Health and Medical Information Needs

Transgender and gender-diverse (TGD) individuals (defined as people whose current gender is different from that assigned at birth, including, but not limited to, nonbinary, queer, and gender nonconforming people, hereafter shortened to *TGD*) face health disparities including high degrees of stigma and discrimination from providers and health care systems [1-4]. Three-fourths of TGD individuals report negative experiences with the health care system [5]. TGD individuals report difficulties finding and accessing TGD-competent health care professionals [6-10], securing insurance coverage for their health care needs [11-13], and finding health care professionals who are sensitive to the needs of the TGD population [14]. Transgender individuals experience stigma and discrimination across the social determinants of health, including bullying in schools, lack of stable income, and quality housing [15-17]. Moreover, TGD individuals often have to manage chronic stress owing to traumatic experiences over their life course [18,19].

Owing to stigma and discrimination when seeking health care, TGD community members often turn to health and medical information on the web [20,21]. Documented examples of insensitive health care include gender insensitivity in which individuals were misgendered (using “he” when a “they” pronoun was requested) or forced care where some patients felt they were forced to do unnecessary examinations or dismissed as “psych cases” [22]. A growing body of literature focuses on the TGD community and their health information–seeking behavior on the web [20,23,24]. A study found that gender transition mental health message boards are popular, especially on the Tumblr platform [25]. In another study, younger TGD individuals used various web-based platforms to explore transgender, nonbinary, and gender-diverse identities and to find support networks [26]. There is evidence that social media is a key resource relied upon by the TGD community to obtain health and medical information [27]. The affordances of social media provide a network for peer-to-peer, emotional, appraisal, and informational support [27].

Considering the known difficulties in seeking and determining the credibility of web-based transgender health information and the current lack of transgender-specific materials on the web [20,24], we aimed to create a health information resource to support the TGD population. For this project, credible information was defined as information created or disseminated by clinicians and organizations, such as the Trevor Project [28], with expert knowledge about care specific to the needs of transgender individuals, and reputable sources of health information such as MEDLINEPlus [29]. Published literature highlights the lack of credible web-based information resources dedicated to the needs of TGD individuals [20,30,31]. Digital tools might be important for TGD health self-management, but they are currently underutilized [32].

To address this need, we developed the Transgender Health Information Resource (TGHIR). We chose a mobile app (Android and iOS mobile operating system) user interface as the optimal method of delivery because of mobile phone ownership—85% of Americans own a smartphone [33]—and mobile phones support anywhere and anytime access to information.

Participatory Design and Web-Based Participatory Design

Participatory design has proven to be successful in designing mobile health resources. The benefits of this approach include engaging co-designers (henceforth, participants) selected to be representative of the community of intended end users to thoroughly explore and prioritize target audience needs [34-36]. Researchers and participants can collaborate and design [37] interfaces that make information accessible. Representation in the design process is helpful because researchers often do not understand how others are affected by technology performance [38].

Leveraging the tacit knowledge and lived experiences [39,40] of individuals from a community helps to understand how everyday tasks are conceptualized, approached, and completed. This process is typically made easier through in-person design sessions. The benefits of in-person collaboration include rapport building [41], a sense of ownership from participants who co-design the system [42], shared values on which design facilitators can build energy within the participatory design methodology [39], as well as perceiving subtle cues of interpersonal communications such as facial expressions and body language [40]. During in-person design sessions, brainstorming and drawing activities are conducted using tangible tools, including butcher paper (a type of heavy paper hung on walls to collect and record ideas) and sticky notes for rearranging the linkages, groupings, and prioritization of specific ideas, allowing for rapid iteration. These activities, and their necessary physical tools, support collaboration and inspire meaningful dialogue between participants and researchers [43].

Approaches to evaluating participatory design have focused on the processes deployed, effects on designers, and outcomes such as satisfaction and empowerment [44,45]. Evaluation of the design process and decision-making have included the collection of qualitative data from end users and system developers to determine the effectiveness of the decision-making [46]. When the focus was on the effect of the design process on participants, evaluators concentrated on participant experiences through interviews [44]. Outcomes, such as participant gains, can also be evaluated, by assessing participant experiences through interviews or surveys to measure how the design product

benefited them and if that benefit lasted [47]. The methodological discipline of participatory design is based on meaningful communication between participants and researchers and allows decision-making by participants, such as how health information is accessed and displayed.

The TGHIR design process coincided with the early months of the COVID-19 pandemic, which forced a transition from in-person to web-based design sessions. Although remote collaboration for design-based user experience interviews was becoming more common [48,49], the use of a suite of web-based tools to support web-based design collaboration has been less documented, especially in the TGD community [50]. Early participatory design studies [50-52] focused on how communication technologies such as email and websites could organize the web-based design process. The effective use of these communication tools, in combination with a shared web-based space hosted on the internet where design could happen synchronously or asynchronously, led to the advancement of how participatory design could be implemented. The addition of shared web-based creative spaces supported participation among remote team members while requiring fewer resources.

Web-based participatory design to support community-driven development of products on an asynchronous discussion forum has proven to be successful. Research conducted by Hess and Pipek [53] indicated that engaging web-based communities to support community-driven development of consumer software is possible, especially if the work is intrinsically fun. The authors found that participants on the web could contribute to the design process of a software system. However, the project began to feel unpaid by some members. A power balance between participants on the web and professional designers was observed and influenced the decisions made for the system. The findings of this study suggested that the responsibilities of participants on the web should be limited to distinct use cases so the development process is not dominated by the most engaged volunteers who might have affordances, such as levels of experience or more time to participate, which may allow greater influence on design decisions. Our proposed web-based participatory design addressed some of these challenges.

Participatory design has been used by other research teams to support the design of health information resources [54-59]. This project was potentially the first to engage TGD participants exclusively in a web-based participatory design process. The web-based method of engagement might safeguard privacy and safety and allow greater involvement of TGD individuals in research. The overall objective of this paper was to describe our web-based participatory methodology and engagement evaluation for design session 1.

Methods

Overview

The setting was the University of Colorado Anschutz Medical Campus, an urban academic medical center, in collaboration with the University of Colorado Integrated Transgender Clinic, and One Colorado, the state's leading advocacy group for LGBTQ (lesbian, gay, bisexual, transgender, and queer) persons. The design and development of the TGHIR were guided by a participatory process to ensure that the final TGHIR design would serve the health needs and goals of the TGD community. The process involved deploying a series of iterative methods to explore the use context and needs of end users. We described and characterized web-based methods and engagement, including qualitative insights from participants on the design of the TGHIR and the number of influential verbal and web-based participant interactions.

Recruitment

There were 3 groups involved in the design of the TGHIR: researchers, advisers, and participants. Researchers were responsible for facilitating the design sessions and implementing the decisions made in partnership with the advisers and participants as well as the agile [60] development of the resource. Advisers were partners [61,62] in the design process and provided feedback on participant engagement strategies, insights from design sessions, and the development process. Participants were responsible for generating ideas and making decisions on how the TGHIR should be designed, features to include, and wireframing.

Participants had to be aged 18 years and meet one of the following two inclusion criteria: (1) self-identified as TGD at any point in their journey or (2) be parents or guardians of a TGD youth.

Recruitment took place during the COVID-19 stay-at-home orders from the State of Colorado. Strategies for recruitment included posting web-based flyers in TGD Facebook groups and delivery through listservs. The outreach was led by a local TGD community partner, One Colorado [63]. We conducted a thorough eligibility call over the Zoom videoconferencing platform (Zoom Video Communications) with each participant to assess their interest in transgender health and how they would like to contribute, which allowed the researcher to confirm the intent of their interest and that the participants could connect to Zoom [64].

Community Advisory Board members, known as advisers, were nominated, after funding for the project was obtained, by partners and allies in the transgender community, One Colorado, and the Integrated Clinic at the CU Anschutz Medical Campus. The process of adviser selection included a nomination phase and an interview phase to assess whether the nominee's goals and motivation fit with the objectives of the project. All nominated individuals were onboarded. Community Advisory Board membership included 8 TGD individuals, 3 parents of TGD adolescents, 1 advocate from One Colorado, 4 health care providers who served the TGD population, 1 library scientist, and 4 research staff, totaling 18 members (3 advisers were in 2 of the categories described above). Advisers attended a 4-hour in-person kickoff meeting and eight 1-hour web-based meetings. The topics of the 8 web-based meetings included purpose-to-practice exercise, identifying credible health information, asynchronous work group planning, a discussion on the Black Lives Matter movement in the summer of 2020, a review of health care provider resources, focus group and design session data discussion, TGHIR app demo feedback,

usability testing data discussion, and a celebration to acknowledge what we achieved together. Each adviser was compensated US $50 per hour for attending the meetings.

Participatory Design Sessions

A web-based design approach required adapting in-person participatory design approaches to understand the targeted end users, the tasks end users were attempting to complete, and the environment in which participants completed the tasks [59]. The three stages of the TGHIR participatory design approach were adapted from Spinuzzi [65] and included (1) initial exploration of end-user needs, (2) discovery processes of prioritization and ideation on potential outcomes, and (3) prototyping. As a research team, we integrated these stages into a larger design process to access the opinions and experiences of participants on behalf of the targeted end users. A total of 4 distinct design sessions were conducted as part of the TGHIR design process (Table 1). We described the methodology implemented in these sessions, including how the methods were adapted to web-based interactions and the degree of participation in the "Ladder of Participation" column [61,62], a theoretical construct that described the range of co-designer participation from low to high.

Several web-based tools were used for web-based participatory design session implementation, including Zoom [64], Mural (Mural) [66], REDCap (Research Electronic Data Capture; Vanderbilt University) [67], and Justinmind (Justinmind) [68]. Zoom was used primarily to allow all participants to meet, with audio and video capabilities using a computer or tablet and computing functionality in a common web-based space. Mural was used as the collaborative workspace for the design sessions; participants were asked to create free accounts and were then sent a link to collaborate in a Mural workspace. REDCap was used for web-based surveys and Justinmind software was used for wireframing and prototyping.

Table 1. Four design sessions.

Design session and goals (research participants)	Participatory design stage adapted from Spinuzzi [65]	Ladder of participation	Web-based tools	Planned in-person tools
Session 1: Exploration of potential features (n=4)	Stage 1: initial exploration of end-user needs	Consultants: participant feedback on features	Mural and Zoom	Butcher paper, sticky notes, markers, and bullseye visualization
Session 2: Feature prioritization (n=22)	Stage 2: prioritization (Kano) and ideation on potential outcomes	Consultants or partnership: participant feedback via Kano and decisions on prioritization	Mural, Zoom, and REDCap[a]	Paper survey, butcher paper, sticky notes, markers, and bullseye visualization
Session 3: Iterative prototyping • 3.1: Wireframing and prototyping (n=4) • 3.2: Account creation (n=4) • 3.3: Aesthetics(n=1)	Stage 3: prototyping	Partnership: participants decide how the resource displays each feature	Mural, Zoom, and Justinmind	Butcher paper, sticky notes, markers, PowerPoint to create first draft screen visuals, prototyping software
Session 4: Usability testing (n=2)	Heuristic evaluation: cognitive walkthrough	N/A[b]	Zoom, REDCap, and the TGHIR[c]	The TGHIR, paper, and pencil

[a]REDCap: Research Electronic Data Capture.

[b]N/A: not applicable.

[c]TGHIR: Transgender Health Information Resource.

Design Session 1: Exploration of Potential Features

We described and characterized engagement in this session and provided findings in the results. Overall, design session 1 included 4 participants who identified as transgender (2/4, 50%) and nonbinary (2/4, 50%). The races reported by participants included American Indian (1/4, 25%), multiple races (1/4, 25%), and White (2/4, 50%). Age ranges were as follows: 20 to 29 years (1/4, 25%) and 30 to 39 years (3/4, 75%). Participants lived in rural areas (1/4, 25%) and urban locations (3/4, 75%).

Design session 1 was exploratory, informed by stage 1 of participatory design approach by Spinuzzi [65] and facilitated by the first author, a member of the research team. The focus was on gathering information from participants about the mobile resources end users in our target audience liked using and what features made their experience with the resource enjoyable. Participants developed personas through an exercise in which they ascribed feelings, values, and behaviors, resulting in potential use cases. An example of a persona statement is provided in Figure 1.

Mural (Figure 2), a web-based collaborative platform, allowed us to use the electronic counterparts of butcher paper, sticky notes, electronic pens with assorted color ink, and distinct colors and sizes of fonts.

We held a 10-minute Mural training session to optimize the time we had with the participants. Owing to the exploratory nature of design session 1, multiple brainstorming activities were conducted. Typically, participants wrote on sticky notes and organized their ideas on a whiteboard. Mural allowed for this functionality. In this session, we asked participants a series of questions about their mobile resource preferences, including, (1) What mobile apps do you like and why, (2) What mobile apps do you dislike and why, (3) What mobile apps do you use often and what makes them reusable, (4) What mobile apps have you stopped using and why, (5) What makes information on a mobile app credible, and (6) What websites do you use to

find health information? Design session 1 also included an activity in which participants placed mobile resource features identified in the brainstorming activity on a bullseye image indicating how important the feature was to them.

Figure 1. Persona statement.

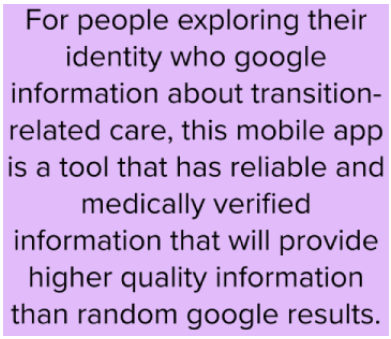
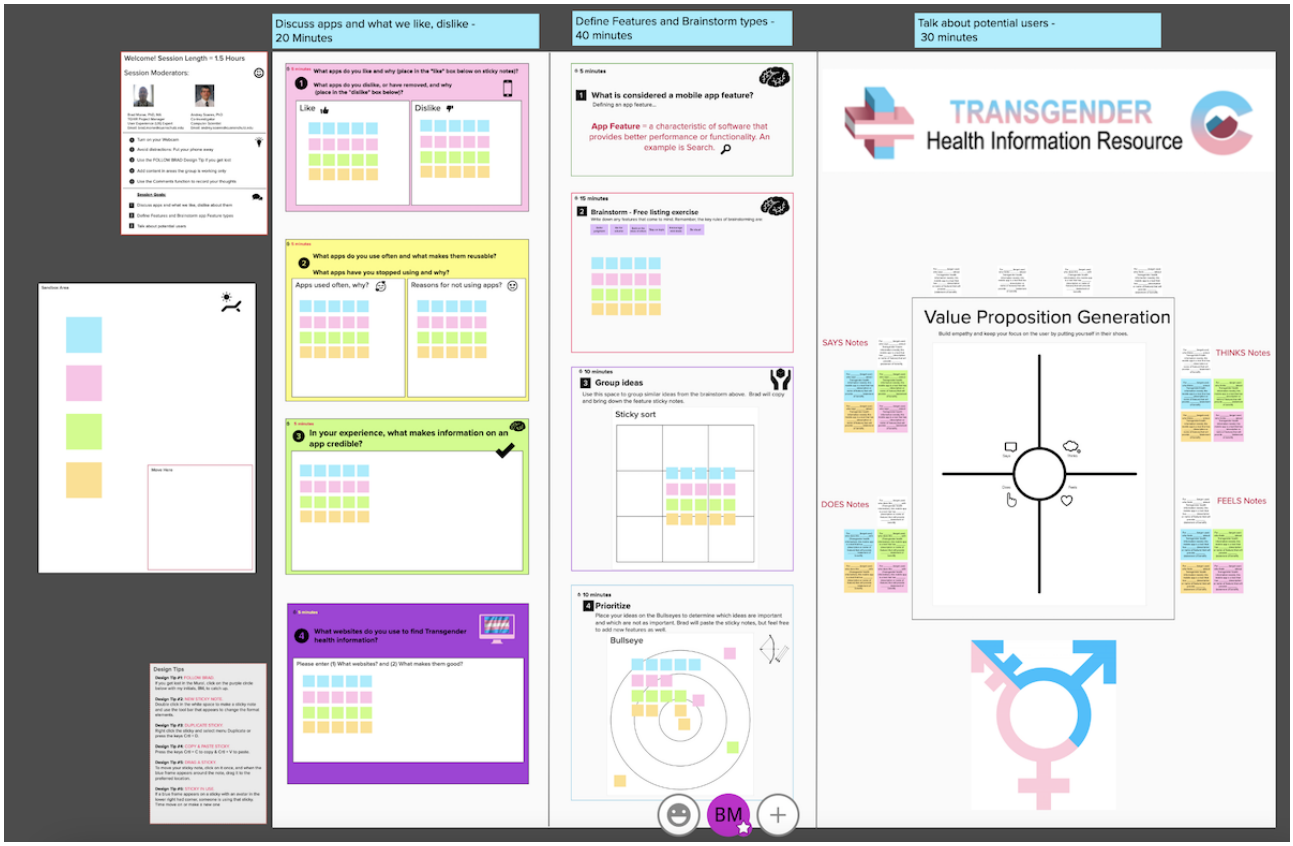


Figure 2. Mural collaborative space.

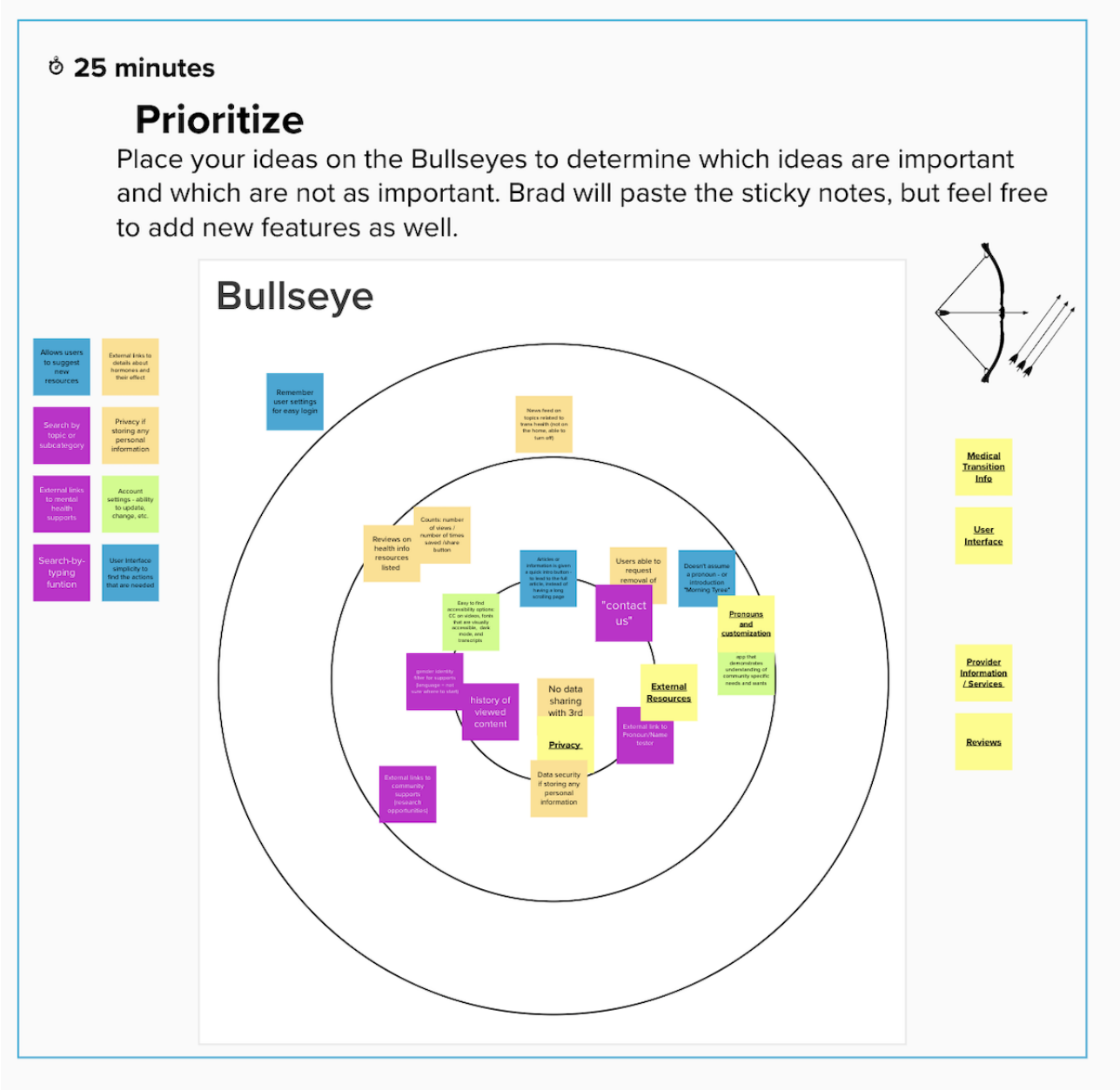


Design Session 2: Feature Prioritization

Design session 2 focused on discovering and prioritizing resource features identified in the first design session using stage 2, the discovery processes, of the participatory design approach [65]. The prioritization of features was organized using a Kano model of customer satisfaction [69-72]. This method assessed participants' opinions regarding a feature being implemented [69]. The transition to web-based implementation required our design team to collect the Kano survey of customer satisfaction through REDCap instead of on paper and in-person. In this exercise, we used Mural to display the features generated by participants in design session 1 so the feature could be prioritized. The participants went through the survey as a group but responded individually.

After the Kano survey was completed, a second round of placing features and categories on a priority bullseye visualization (Figure 3) was conducted as a design exercise, furthering the discussion on what features were the most desirable to end users and should therefore be developed first. The design session 2 bullseye exercise was compared with how features were prioritized using the Kano survey. In addition, this exercise allowed participants to group the features into categories, such as in-person card sort, by grouping different sticky notes that referred to similar or the same type of features [73,74]. The last activity in design session 2 was the initial prototyping of the home menu and health resource preview, which was performed in Mural.

Figure 3. Bullseye design exercise.



Design Session 3: Iterative Prototyping

Design session 3.1 and design session 3.2 focused on wireframing, a process in which a sketch is made of what a product, in this case, a user interface, may look like. The wireframe was used as a starting point for the design work [75] and prototyping of the health information resource as an intentional and planned health information–seeking experience in stage 3 of the participatory design approach [65]. This work was performed using the Justinmind tool, a program that allows designers to rapidly create interfaces and modify them in real time. Using Justinmind, we created wireframes and asked participants for input on the design and implied functionality using Zoom. Participants first responded to the design elements of the baseline design created by the researchers. In traditional in-person sessions, the pencil and paper design methodology allowed for a quick iteration of the initial design during an in-person session. In this web-based design session, the use of a baseline design allowed the researcher to engage more quickly and in greater detail about the interface because time was not used to set up the initial design. Design session 3.3 was held with one participant to collect insights into aesthetics, or look and feel, of the TGHIR.

Design Session 4: Usability Goal-Oriented

The fourth and final design session occurred after the TGHIR was built and consisted of a cognitive walkthrough heuristic evaluation, in which an experienced mobile health researcher and a behavioral scientist were asked to perform navigationally based tasks, assuming that most tasks embedded in the TGHIR would be goal-orientated [76], for example, find and like a resource. Participants installed the resource on their phones for an authentic experience.

In the usability testing sessions, Zoom, REDCap, and TGHIR were used. In our initial plans, this interaction would have been in-person and the participant would have had the TGHIR in

their hands, as we observed their use of the resource and recorded the necessary usability data. Instead, design team members observed the participants through a Zoom connection. Usability evaluation tasks (Table 2) were described and completed by the participants [77,78]. These tasks were prioritized because they were associated with the features participants identified as important for the TGHIR to access credible health information.

Table 2. Evaluation tasks.

Task	Action to be completed
1	Create account and view consent and privacy language
2	Select preferred health information categories
3	Find a specific health information item using the category cards
4	Find and use the filter to narrow the resources to a relevant informational item
5	Like a resource
6	Bookmark a resource
7	Locate and use the search function to find a specific health information resource
8	Send a message to the developers
9	Share a new resource for the community with the developers
10	Find the most liked health information resource

Data Analysis

We analyzed the video recording from design session 1 and the artifact created in the Mural collaborative space to describe and characterize web-based engagement. Design session 1 was recorded using the Zoom tool and transcribed by a professional transcriptionist. A research team member performed rapid analysis [79] of the transcriptions to quickly identify key points and comments that reflected participant engagement and major design decisions. Exemplar quotes were provided to highlight the impact of comments on the final design and the resulting features developed in the resource. Quantitative data included length of each design session exercise, number of verbal comments by all participants, total verbal comments, number of times the Zoom camera was turned off by participants, number of Mural interactions (creation or manipulation of digital Mural content) by participants, and total Mural interactions by design exercise. For a verbal comment or Mural interaction to be counted, the comment or interaction had to be considered influential in advancing the design process. Comments or interactions that expressed confirmation or agreement were not considered influential.

Ethics Approval

Informed verbal consent was obtained from all participants. Each participant was paid US $50 for each of the 1.5-hour design sessions they attended. Project approval was obtained from the University of Colorado's Human Research Ethics Committee, Colorado Multiple Institutional Board (COMIRB# 19-1562).

Results

Recruitment

Eligibility screening was performed using Zoom. A total of 41 individuals were screened for eligibility; of these, 27 (66%) were eligible and available to participate in at least one of the design sessions. Mural relied on well-established computing conventions to add, edit, and delete content and was, therefore, familiar to the participants who quickly mastered the skills to work alongside the research staff in the collaborative space.

Design Session 1

In design session 1, participants identified the following specific health information for inclusion in the resource: affirming care (supportive care for the TGD community [7]), affordable medical options, information on transition, information on successful transitions (transitions were associated with the period during which a person began to live according to their gender identity rather than the gender they were thought to be at birth [80]), family resources, community support, and medically verified information.

Responses to the questions addressing participants' mobile app preferences are presented in Table 3.

We provided the number of verbal comments and web-based interactions in design session 1 to show the volume of participant engagement in Table 4. Design session 1 was 119-minutes long; there was an average of 1.87 web-based points of engagement every minute. The researchers turned off their cameras to focus on the participants. Two participants experienced bandwidth trouble during the first exercise and chose to turn off their cameras to contribute to the web-based participatory design process. These 2 participants did not provide any other reason for their cameras being turned off and both continued to contribute despite the bandwidth problems they experienced.

Additional qualitative evidence for the effectiveness of the web-based participatory design process is presented in Table 5. Participant insights from the first design session remained prominent throughout the design process and directly affected the course of the design work. Participant's verbal interactions were evident in the final TGHIR features.

Table 3. Questions addressing participant's mobile resource feature preferences.

Question	Responses
What mobile apps do you like and why?	• Gmail: simple design, easy to access, can limit notifications • Facebook: easy to refresh • Uber • Reddit: the ability to search by subcategories • Spotify • Genius Scan • Evernote: does a fair job of talk to text • Merlin Bird ID: simple, easy to use, and regularly updated • Mint: great UI[a] and UX[b] and never buggy • Native Land: great multisource database and simple UI and UX • AllTrails: surprisingly prefer using mobile over web app because of UI and UX
What mobile apps do you dislike and why?	• Whole Foods: not intuitive • Snapchat: too many notifications • Your Turn: advertisements interrupting experience • Tabletopia: optimized for PC and Tablet not mobile • Spectrum Mobile: lack of control with certain items • C25K: too many options • Stitcher: too many options • Apple Notes app: clunky and difficult to change formats • Google sheets: difficult to use • Apple Maps: location and information often totally inaccurate
What mobile apps do you use often and what makes them reusable?	• Hearthstone: rewards frequent use • Facebook: allows me to keep connected • Reddit: plenty of media available • Discord: able to access through multiple mediums • Smarthub: saves relevant data for later • Keep Notes: does exactly what it needs • Instagram: addictive, friends, distraction • Stitcher: do not like the interface, I just like the content, streaming, and downloading • NYTimes: good usability • Gmail: necessity and like UX better than other mail apps • Spotify: seamless desktop to mobile transition
Reasons for not using an app	• Excessive or random crashes • Too many ads • Excessive notifications • Difficult to navigate; no search function • Too many options • Need to create an account when not necessary • Ugly or outdated • Uses too much power or memory on phone • Busy user interface
What makes information on a mobile app credible?	• Citing sources • Current information • Customer service • Association with credible groups • Not asking to rate the app • Easy to navigate • Inclusive language • Transparency on who developed app
What websites do you use to find health information?	• Private Facebook groups • Forums like Reddit • WebMD • Denver Health • PFLAG • One Colorado • Queer Asterisk

[a]UI: user interface.

[b]UX: user experience.

Table 4. Design session 1 engagement evaluation (total interactions, N=301).

Mural exercise	Exercise time (length of discussion)	Verbal comments by participants, n				Verbal comments per exercise, n	Web-based interactions by participants, n				Web-based interactions per exercise, n
		A[a]	B[a]	C[a]	D[a]		A[a]	B[a]	C[a]	D[a]	
What apps do you like	6:10-14:50	2	2	0	1	5	8	10	3	7	28
What apps do you use	14:51-22:29	1	1	3	3	8	11	10	7	10	38
Credible information	22:30-30:30	5	3	0	1	9	5	5	4	4	18
TGD[b] information	30:31-39:05	1	2	0	0	3	5	4	4	2	15
Brainstorm—free listing	39:06-1:00:38	8	7	1	3	19	10	8	9	8	35
Card sort	1:00:39-1:13:24	3	5	2	2	12	12	14	10	10	46
Bullseye prioritization	1:13:25-1:28:00	7	7	3	3	20	8	12	8	0	28
Value proposition generation	1:34:05-1:55:10	2	1	0	0	3	4	4	2	4	14
Total	1:58:55 (including breaks)	29	28	9	13	79	63	67	47	45	222

[a]Participants.

[b]TGD: transgender and gender-diverse.

Table 5. Exemplar quotes, impact, and design influence.

Session Exercise	Result		
	Comment	Impact on design	Influence on design
1.3	"I think having a channel in which folks can reach out if there's an issue with information on the app...message customer support, call customer support or some kind of way that it's not just, you know, you're not searching really, you know, a long way through the app or trying to find a human to connect with within even an issue or a question"	Customer support as a priority within the app—visible, easily accessible. Important to make users feel valued and heard.	Contact Us page with the option of sending different types of messages to the TGHIR[a] development team.
1.5	"I think it would be good if there is a place for it [the resource] to remember your name but also not to make it too cumbersome should you decide to change it. If it remembers your names also having an option for pronouns, even if it doesn't pop up anywhere, it's nice to have that affirmation."	Function to remember names and customize pronouns within the resource. Demonstrates understanding of target audience.	Providing pronouns was eventually scrapped to avoid collecting data the resource would not use. This is the settings menu based on the comment above. The design attempts to make changing settings easy.
1.5	"My Verizon [app] can update some things on your account, but otherwise it's just a giant ad for you to upgrade your system. I think interactivity, there's a reason why you've downloaded an app instead of going to the website."	Information should be presented in a mobile app that fosters easy access with an intuitive interactive navigation so the app could be used at any time and in any place.	The TGHIR was design and developed for iOS and Android, the 2 most common mobile app operating systems.
1.6	"I have the [Name], My Chart open. And one of the options is you can personalize it. The personalization is just changing the color for the frame of the app. That seems pointless to me...yeah, it just seems like such a superfluous option. I would rather move the icons around on the homepage...there might be something that I like to check more than other people. Moving the menu around would be a nice personalization. But changing the color is not worth it."	If the resource is going to include customization options, it should go beyond the ability to simply change the colors on the interface, eg, the option to move icons around based on relevancy to the individual user. The orange cards are categories of interest selected by the user.	The Health Resource Cards could be pinned to the top of the page making them easier to return to during repeated use. Once the resource was pinned, the TGHIR app changes the card from blue to orange.

[a]TGHIR: Transgender Health Information Resource.

Discussion

Principal Findings

We found that using a web-based suite of collaborative tools with participants, we were able to effectively engage in productive discussions and make design decisions for the development of TGHIR. This approach to design work, in our case, was seamless and did not limit participants from engaging and providing meaningful influence on the resulting health information resources. The value of web-based recruitment and design sessions should be underscored because of the relative social safety of TGD participants. This recruitment method may be a strength for engagement and have a positive impact on TGD involvement in research by creating safe web-based spaces for TGD involvement.

We observed consistent engagement throughout design session 1. Upon reflection and analysis of the data, we found that the immediacy of engagement was impressive. It did not take participants much time to get involved and discuss design issues with their fellow participants. The dual interaction of manipulating web-based content in the Mural document and discussing the topic at hand may have led to a more immediate collaboration.

This study shed light on a web-based methodological approach to co-design health information resources within TGD communities. Through their involvement and enthusiasm for the work, participants indicated that a web-based approach to design was appropriate and can be used instead of resource-intensive in-person gatherings. Queer and trans communities have embraced digital technologies in radically affirming ways [81,82] to move beyond the acceptance of unsatisfactory options. The uptake of web-based design, and the necessary digital technologies, in TGD communities is feasible.

Mural interactions indicated that participants were consistently present throughout the design process. With 222 Mural interactions across 4 participants, it was evident that these individuals were highly engaged on the process. Whether this level of engagement can be replicated or generalized to other communities and topics requires further research. The verbal comments provided evidence that the interactions were process-oriented and moved the discussion to inform the development of TGHIR. Furthermore, specific design decisions were made owing to the input of the participants, providing evidence of their importance in the web-based design process and speaking to the effectiveness of web-based participatory design.

Strengths, Limitations, and Future Directions

The strength of this project was the introduction of a method to describe and characterize engagement and interactions during participatory design facilitated by web-based means. The first limitation was that this study evaluated the engagement of participants from the TGD community but did not empirically compare the quality or quantity of engagement with an in-person design process. Future research should examine in-person design sessions compared with web-based design sessions to evaluate and compare the quality and quantity of engagement and interactions. Second, our recruitment method used Facebook to identify participants. This approach yielded a predominantly educated, professional, urban, and white sample. Although we recruited enough TGD individuals for the design of the TGHIR, we acknowledge that this sample may not have been representative of the overall population. As a result, we sought and were awarded funding to test the resources using a diverse research sample. This study is currently underway. Third, the small number of participants was a limitation. In future work, more participants should be involved in the participatory design process to determine whether high levels of web-based participation are maintained in larger samples. Finally, although web-based means of participation helped during the COVID-19 pandemic, a limitation was that it might have also been difficult to reach individuals who did not have the means or ability to access the requisite resources for web-based collaboration, such as high-speed internet and private space [83]. It will be important to keep this accessibility issue in mind when recruiting participants.

Conclusions

Our results had important implications for the use of web-based methodologies in the design of health information resources. Web-based participatory design can support opportunities to contribute despite the potential logistical barriers of in-person design sessions by offering multiple convenient design session times and multiple interaction options. In addition, this approach is helpful when recruiting members from marginalized communities that are small and geographically dispersed, especially rural communities. Not only does the web-based methodological approach work during a pandemic but it may also help when there is historic distrust of research and health care from a community that has been stigmatized and experienced discrimination, such as the TGD community, by researchers and medical providers.

Our evaluation of the web-based participatory design indicated that web-based design sessions can engage participants in creating satisfactory interfaces for accessing and consuming health and medical information. Obtaining web-based input from participants was possible and efficient. Web-based recruitment is also possible for individuals who belong to marginalized communities and provides a platform through which these individuals can safely communicate with others in their community to design health information resources. Integrating web-based platforms can effectively engage participants and yield a positive user experience. Multiple participants reported that a health information resource of this nature would have been helpful in their journey toward gender identity exploration or gender transition.

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Data Availability

Relevant data associated with, created, and used for the evaluation presented in this paper are openly available from the University of Colorado Strauss Health Sciences Library, Anschutz Medical Campus, Data Repository [84].

Authors' Contributions

BM was responsible for designing and facilitating the participatory design process, coding the qualitative manuscript and web-based interactions, and served as the lead author. AS was responsible for designing the methodology, agile development of the resource, and served as the second author. KY was responsible for obtaining data in usability tests, coding the qualitative transcript and web-based interactions, and reviewing and editing the manuscript. KD was responsible for cataloging the content in the resource, designing the search for credible transgender health resources, and for reviewing and editing the manuscript. MA was responsible for the design of the methodology, the interpretation of data, and for reviewing and editing the manuscript. BDH was responsible for designing the qualitative methodology for focus groups, interpretation of focus group data, and reviewing and editing the manuscript. RSL was responsible for designing the methodology, interpretation of the data, and for reviewing and editing the manuscript. BMK was responsible for designing the methodology, interpretation of the data, and for reviewing and editing the manuscript. LMS was responsible for designing the methodology, interpretation of the data, for reviewing and editing the manuscript, and served as the senior author.

Conflicts of Interest

None declared.

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Abbreviations

LGBTQ: lesbian, gay, bisexual, transgender, and queer
REDCap: Research Electronic Data Capture
TGD: transgender and gender-diverse
TGHIR: Transgender Health Information Resource

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Original Paper

Development of a Family-Centered Communication Tool for Kidney Health in Premature Infants: Qualitative Focus Group Study Using Human-Centered Design Methodology

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Abstract

Background: Premature infants are at increased risk of kidney-related complications, including acute kidney injury (AKI) and chronic kidney disease (CKD). The risk of CKD in prematurely born infants is underrecognized by health care teams and caregivers. Understanding how to communicate the risk of CKD to caregivers is essential for longitudinal clinical follow-up and adherence.

Objective: This study aimed to determine family caregiver attitudes toward kidney health and risk communication during a neonatal intensive care admission. We also sought to understand caregiver preferences for the communication of information surrounding the risk of CKD in premature infants.

Methods: We augmented standard qualitative group sessions with human-centered design methods to assess parent preferences and clinician perspectives. Caregivers recruited had a prematurely born child who spent time in the neonatal intensive care unit at Riley Hospital for Children in Indianapolis, Indiana, and experienced AKI or another kidney complication, which put them at risk for future CKD. We used a variety of specific design methods in these sessions, including card sorting, projective methods, experience mapping, and constructive methods.

Results: A total of 7 clinicians and 8 caregivers participated in 3 group sessions. Caregivers and clinicians readily acknowledged barriers to and drivers of long-term kidney monitoring as well as opportunities for communication of the risk of long-term kidney disease. Caregivers' primary concerns were for both the type and depth of information conveyed as well as the time at which it was communicated. Participants emphasized the importance of collaboration between the hospital care team and the primary care provider. Participant input was synthesized into several prototype concepts and, ultimately, into a rough prototype of a website and an informational flyer.

Conclusions: Caregivers of premature infants are open to communication about kidney health during their neonatal admission. The next phase of this work will translate caregivers' preferences into family-centered communication tools and test their efficacy in the neonatal intensive care unit.

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KEYWORDS

qualitative research; patient-reported outcomes; neonates; chronic kidney disease; human-centered design; acute kidney injury; kidney health

Introduction

Premature infants are at high risk of kidney-related complications, including acute kidney injury (AKI) and chronic kidney disease (CKD) [1]. AKI is common in premature infants, occurring in between 20% and 40% of infants, depending on the patient population studied [2,3]. Premature infants with AKI have higher rates of mortality and longer hospital stays [2,3]. The risk of kidney-related complications in premature infants does not disappear after the neonatal admission. Studies in prematurely born children show a 4-fold increase in CKD during childhood and adolescence [4-9]. While likely multifactorial, one explanation for this increased CKD risk is that premature infants are born with a decreased number of nephrons due to their early delivery [10]. Furthermore, the extrauterine environment (including the use of nephrotoxic medications and perinatal stressors) may not be amenable to proper nephron development [11]. Even children with normal kidney function but a history of AKI have a 10 times higher risk of developing kidney failure before the age of 40 years [12,13]. Thus, as more critically ill infants survive and live into adulthood, the impact of kidney health on premature infants is a significant long-term concern.

Communication surrounding kidney health to families, specifically focusing on the risk of CKD, is essential in empowering families and ensuring longitudinal clinical follow-up and monitoring. Studies show that kidney health, including the diagnosis of AKI and the risk of CKD in prematurely born infants, is underrecognized by health care teams [14,15]. While there are no established best practices for communication in the neonatal intensive care unit (NICU), families of premature infants report a desire for direct and concise communication during their NICU stay, focusing on the most urgent or immediate clinical concerns [16,17].

There have been no studies which evaluate kidney-specific health communication with families. The purpose of this study was to fill this gap by evaluating caregiver attitudes toward kidney health and CKD risk communication as well as caregiver preferences for communication of information surrounding the risk of CKD. This study serves as the first step in the development of a family-centered tool to improve communication about kidney health in premature infants.

Methods

Overall Approach

In collaboration with Research Jam and the Indiana Clinical and Translational Science Institute's Patient Engagement Core, we conducted 2 phases of group sessions using qualitative focus group methodology augmented by human-centered design methods (Multimedia Appendix 1). Human-centered design, which is increasingly used within health care, is an iterative design process where stakeholders most closely affected by the problem or solution are engaged in developing the solution [18,19].

Sessions were facilitated by 4 research specialists using human-centered design research methods. Sessions were held virtually through Zoom (Zoom Video Communications), lasted approximately 120 minutes each, and were recorded and transcribed for analysis. All sessions used activities to engage clinicians and caregivers to better understand caregiver perspectives on communication surrounding kidney disease as a first step in the co-design of a kidney disease communication tool [20,21]. Activities were open-ended, allowing for a wide range of responses to minimize bias and for families to be as open and truthful as possible about their preferences. Sessions began with warm-up activities to encourage participation and collaboration [22]. We then used specific generative activities (eg, empathy mapping, detailed below) designed to encourage study participants to express their thoughts and feelings and constructive methods to help with concept development [21]. All sessions used Miro Whiteboard (Miro) [23], a collaborative whiteboard platform which the group facilitator used to document and visualize responses in real-time for the group.

Recruitment, Subjects, and Study Setting

Stakeholders included clinicians and caregivers. Clinicians were from across the United States and cared for prematurely born children who spent time in the NICU. This included physicians, nurses, and nurse practitioners trained in pediatric nephrology, general pediatrics, and perinatal and neonatal medicine, all of whom were approached and recruited by the principal investigator.

Caregivers were recruited who had prematurely born children who spent time in the NICU at Riley Hospital for Children in Indianapolis, Indiana. Caregivers were approached for enrollment in this qualitative study if their child was: (1) born prematurely and admitted to the NICU during their infancy; or (2) experienced AKI or another kidney complication (such as a slow to normalize serum creatinine), which put them at risk for future CKD. Caregivers were eligible for this study if their child was between ages 2 and 25 years old, if they agreed to participate in the web-based session, and if they had no diagnosed cognitive disabilities.

Recruitment was conducted by phone as well as in the outpatient pediatric nephrology clinic at Riley Hospital for Children, part of Indiana University Health, in Indianapolis, Indiana. Permission to approach the caregiver was obtained from the nephrologist of record to ensure the child did not have any medical treatments or conditions that could deter participation in the session. Informed consent was obtained from each study participant. Study participants were given a US $100 Amazon gift card for their engagement.

Exploring and Co-Design

We held 2 virtual sessions that were identical in purpose and methods but engaged different stakeholder groups. The first session included clinicians, and the second session included caregivers (Multimedia Appendix 1).

Specific activities included during the exploring and co-design sessions included:

Empathy Mapping

Empathy mapping is a generative method in which stakeholders are asked to intentionally speak about different aspects of an

experience (thinking and feeling, hearing, seeing, and saying and doing) [24]. Stakeholders (clinicians and caregivers) were asked to address each of these areas based on the following prompt: "After their child has received life-saving drugs in the NICU, parents are told that their child will need lifelong kidney monitoring. Help us understand this conversation." To understand the context, stakeholders were also asked to describe where, when, and how this conversation took place. In addition, stakeholders were asked about the barriers to and drivers of lifelong kidney monitoring.

Co-Design

Co-design refers to the practice of guiding caregiver and clinician co-designers in the design development process [25]. The following co-design methods were used:

1. *Concept generator:* we created a worksheet in Miro to help caregiver and clinician co-designers diverge and converge on the function and form of a potential tool. It included the following instructions:

   We need to develop a tool to help patients and their families overcome their barriers to long-term kidney monitoring. Let's think creatively about what that tool could be.

1. *Prototyping:* creating a rough version of a solution (a prototype) gave designers and caregiver and clinician co-designers the opportunity to make rough ideas tangible to quickly gain feedback and make iterations. Prototypes displayed the approximates of the solution or part of the solution [26]. How the prototype looked at this stage was less important than the conversation about why features were included and what problems each feature solved. We created a worksheet in Miro to help stakeholders create their prototypes.
2. *Rose, Thorn, Bud:* Rose, Thorn, Bud was a reflective activity used during the session to get stakeholders to intentionally think about each prototype and provide feedback [27]. As a group, stakeholders focused on 1 prototype at a time and then shared 3 things: something that they thought was working well (a rose), something that presented a challenge (a thorn), and something that represented an opportunity or idea with potential (a bud).

Analysis of Exploring and Co-Design Sessions

Data (including the developed products, notes, and transcripts) from the sessions were analyzed using John Kolko's methods of analysis and synthesis, using a creative process to connect research insights with design patterns to generate well-grounded design ideas [28]. These data were grouped by affinity or similarity of content, with each group given a heading to summarize its content. The resulting affinity diagrams spatially organized the data into groups based on similarity of content and represented the full picture of the data organized by theme [29]. Next, an analysis team created visual models of the themes and how they were interrelated [28].

Models included a refined empathy map, a communication opportunities map, and a grouping of "must have," "can't have," and "nice to have" features for the communication tool. During model-building, a total of 2 "must have" and "nice to have" continuums were created. Each of the educational content and bonus feature items were placed on their respective continuums as determined by participants. Discussion points collected during the sessions were placed below their related item in the continuum.

Prototype Development

The research team then moved to prototype development, which looked at the outcomes of analysis ("what is") to build solutions for the future ("what could be") using the following synthesis methods:

Brainstorming Potential Challenges to Solve

To diverge further on what the solution could be, the team identified underlying challenges within the main objective. Asking "how might we..." allowed the research team to think beyond first instinct responses and use a divergent mindset to come up with many potential ideas for solutions. The research team then converged on the challenges that best fit the objective and what was learned from the analysis.

Brainstorming Potential Solutions for Selected Challenges

The research team asked one "how might we..." question at a time and listed as many solutions as they could. The research team used a divergent mindset, limited judgement, and focused on quantity over quality. Thinking broadly allowed for the generation of out-of-the box solutions that could be examined for valuable elements that could be implemented into a final tool.

SCAMPER Method to Diverge on Additional Solutions

To further diverge, the research team used the SCAMPER method to create new solutions by manipulating already-stated solutions [30]:

- Substitute: what could you substitute or change?
- Combine: could two or more ideas or pieces be combined into something else?
- Adapt: what could be tweaked to improve the solution?
- Modify: could some solutions be changed to be improved?
- Put to another use: could solutions apply to another use?
- Eliminate: what could we take away from these solutions to improve them?
- Reverse: would rearranging elements improve solutions?

The research team used each of these prompts to create new solutions based on the existing process or solutions from the previous step. Following the divergent stage, the research team reviewed the list of solution ideas and voted for those they thought were the most appropriate and interesting.

Prototyping

The research team individually created prototypes of the tool inspired by the converged list of solution ideas, allowing the research team to explore additional ideas that could be included in the final tool. Refined prototypes were then used to get feedback from the stakeholders.

Prototype Refinement

We held 1 virtual session with a subgroup of clinicians and caregivers by Zoom to evaluate the prototypes developed using Miro.

1. *Sorting to Prioritize Prototype Content and Features:* Study participants were presented with a list of potential educational and informational elements identified as either "must have" or "nice to have" by the research team. Study participants were then asked to discuss and sort each of these into one of the two categories themselves. This same approach was taken with a list of bonus features the tool could include. This activity allowed for potential elements to be categorized based on the perspectives of the stakeholders, not just the research team.
2. *Prototype Feedback using Rose, Thorn, Bud:* Study participants were shown 2 prototypes. Each prototype had 3 main elements: information and education, bonus help, and appointment reminders. Prototype A focused primarily on digital solutions, while prototype B focused on analog solutions. The research team presented both prototypes to the study participants, asked for clarifying questions, then worked through the same Rose, Thorn, Bud activity used in phase 1 to get feedback for each prototype. This activity helped the research team understand elements of the prototypes that stakeholders liked and disliked.
3. *Frankenstein Prototypes:* With knowledge and opinions about what should go into the tool, study participants were asked to build new prototypes using their favorite elements from prototypes A and B. With the ability to mix, match, and create new elements, the research team could see what the participants prioritized.

Analysis of Prototype Refinement

We used affinity diagramming to group the feedback provided during Rose, Thorn, Bud. Through discussion within the research team, feedback from participants was arranged into groups and given thematic headings. These headings were used to identify key elements that study participants liked, did not like, and saw as having potential in the prototypes presented to them, allowing the research team to make final decisions about how to refine the prototypes. The research team then reviewed each item on the continuum and made decisions about what should be included in the final communication tool. Decisions were made based on feasibility and how well the item would address the original objective.

Ethics Approval

This study was approved by Indiana University's institutional review board (protocol #11958), by whom it was deemed minimally risky.

Results

Participants

The exploring and co-design sessions included 15 participants (7 clinicians and 8 parents), while the prototype refinement session included 10 participants (6 clinicians and 4 caregivers). We approached 20 clinicians (7/20, 35% participation rate) and 32 caregivers (8/32, 25% participation rate; Multimedia Appendix 1). All the patients represented by caregivers in this study were discharged from the hospital and were currently seeing pediatric nephrology for monitoring of kidney health or management of CKD. See Table 1 for demographic characteristics for the study participants in the exploring and co-design sessions.

Table 1. Demographic characteristics for study participants in the exploring and co-design sessions.

	Clinicians, N=7	Caregivers, N=8
Gender, n (%)		
Female	4 (57)	6 (75)
Male	3 (43)	2 (25)
Age (in years), n (%)		
21-44	6(86)	7 (88)
45-64	1 (14)	1 (12)
65 and older	0	0
Race, n (%)		
Asian	1 (14)	1 (12)
Black or African American	1 (14)	1 (12)
White	5 (72)	6 (75)
Ethnicity, n (%)		
Hispanic or Latino	1 (14)	2 (25)
Not Hispanic or Latino	6 (86)	6 (75)
Clinical subspecialty		
General pediatrics	1 (14)	N/A
Neonatal and perinatal medicine	2 (28)	N/A[a]
Pediatric nephrology	4 (58)	N/A
Child's current age (in years), mean (SD)	N/A	6 (4)

[a]N/A: not applicable.

Caregiver Experience

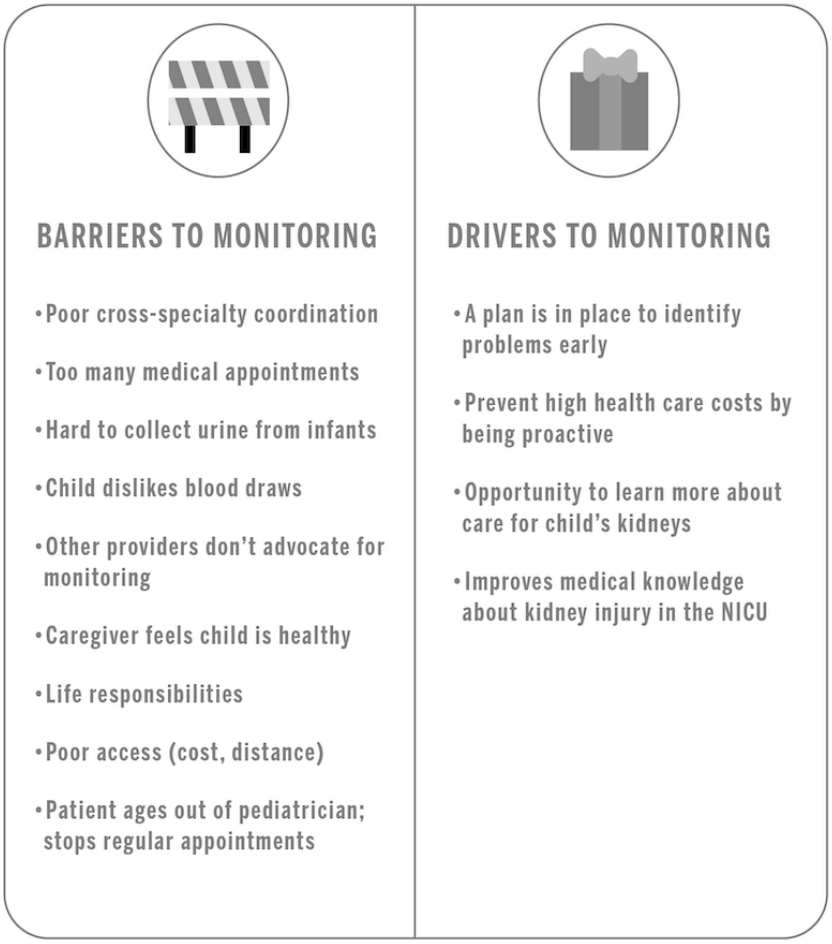
The caregiver experience began with their infant's admission to the NICU. Sometimes, caregivers and clinicians expected that an infant would require immediate medical intervention after birth, while other times it was unexpected. Either way, infants required medical care in the NICU, with their caregiver as the primary decision maker. About this moment, one participant said (paraphrased): "I sat and looked at this perfect baby and they're telling us she has all these challenges." During the course of medical care, parents were often involved in difficult decisions or treatment decisions, such as clinicians recommending the use of life-saving medications and treatments that could harm their kidneys (eg, nephrotoxic medications, surgery, and other interventions; Multimedia Appendix 2).

In addition to making decisions critical to their infant's care, the physical location of the decision placed additional stress on caregivers. Frequently, discussions between caregivers and clinicians occurred in the NICU, sometimes privately but often in the proximity of other patients and passersby. Caregivers described an overwhelming scene with many new sights and sounds, hopes and fears, high and low emotional points, and advanced levels of stress and fatigue. As doctors presented the treatment options and the implications of those options, caregivers found it easy to lose focus and not remember all the details of the conversation. They may or may not remember being informed that the child would need lifelong kidney monitoring due to potential kidney damage from life-saving treatments (Multimedia Appendix 2).

Barriers and Drivers to Monitoring

Some caregivers recalled that clinicians suggested the need for kidney monitoring at the time of discharge. Both caregivers and clinicians readily acknowledged barriers to and drivers of long-term kidney monitoring. Caregivers shared that many of their pediatricians and other health care clinicians agreed with or reinforced the need to monitor the patient's kidneys; however, at least one caregiver was told that it was not necessary by their pediatrician. Adherence to kidney health monitoring, in addition to other treatments that may be required following their NICU admission, posed more immediate challenges, such as the difficulty of their young child tolerating a blood draw or urine collection. Caregivers weighed these barriers versus the drivers of early identification of kidney problems, improved care for their child, and saving money over time. Barriers to and drivers of long-term kidney monitoring are summarized in Figure 1 and Multimedia Appendix 2.

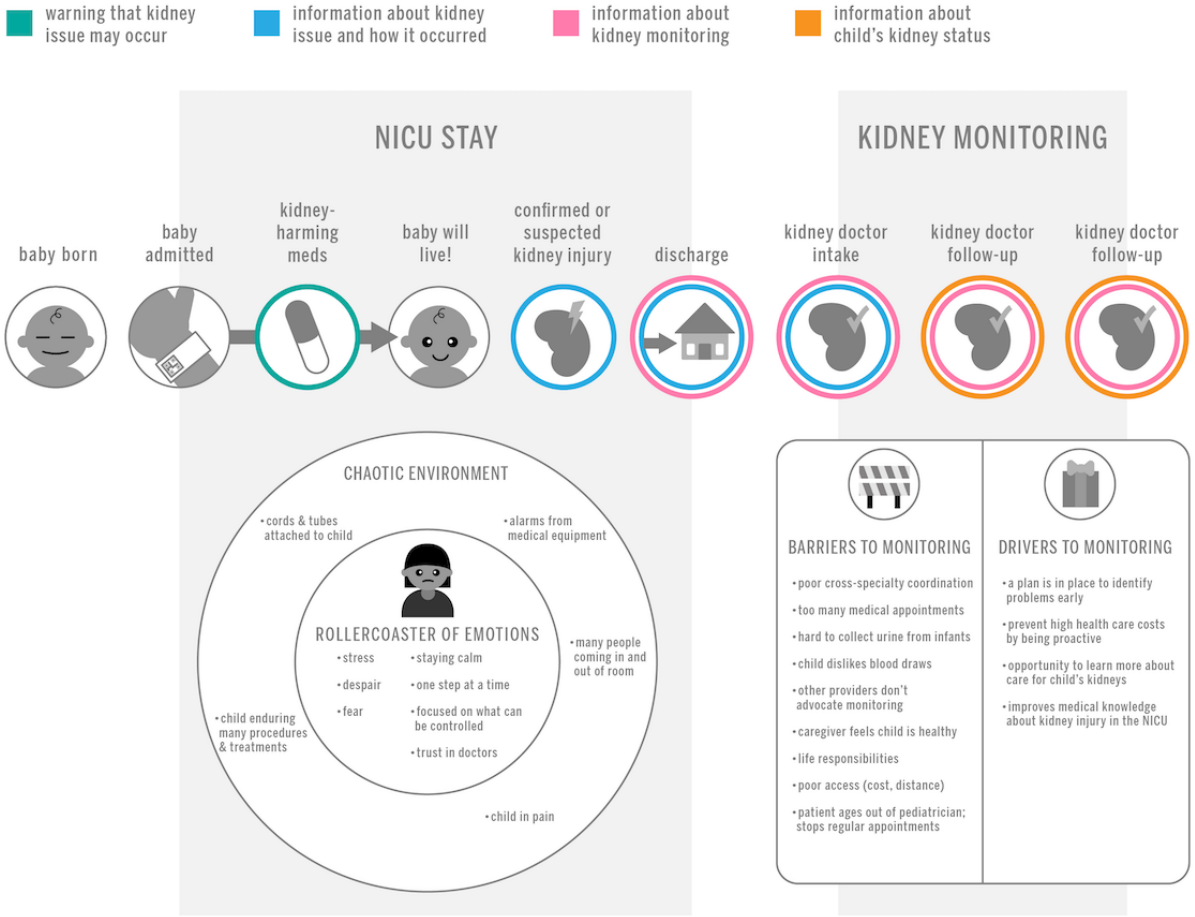
Figure 1. Barriers and drivers to long-term kidney monitoring for children after the neonatal intensive care unit (NICU).



Communication Considerations and Opportunities

Stakeholders reported positive and negative aspects of the communication of medical information, both generally and about the implications of kidney injury specifically (Multimedia Appendix 2). Caregivers noted that due to the stressors experienced by caregivers and the challenges of learning and memory retention in the NICU, clinicians should offer information about kidney health and long-term kidney monitoring at multiple points throughout the NICU admission, including at the time of administering medications or therapies that may contribute to kidney injury, at discharge as part of the discussion of follow-up care needed, and at follow-up appointments. Figure 2 shows a model of caregiver experience with communication opportunities identified.

Figure 2. Communication opportunities for kidney monitoring during and after neonatal intensive care unit (NICU) stay.



Contents and Features

Study participants sorted educational content and potential features into "must have" and "nice to have" categories. Each of the educational contents was placed on their respective continuums based on where it was placed by study participants (Table 2). For example, "questions to ask clinicians" was placed in the "must have" section of the education and information continuum because both groups sorted it as a "must have," while "NICU guide" was placed both in the "must have" section and the "nice to have" section because 1 group sorted it as a "must have" item and the other as a "nice to have item."

Table 2. "Must have," "nice to have," and "should not have" components determined by caregivers and clinicians.

	Must have	Nice to have	Should not have
Educating caregivers and patients	• Kidney condition treatment and monitoring • Tips for lifelong monitoring • Real-world experiences • Benefits of monitoring • Appropriate tone • Presented in simple terms • Use visuals	• Real-world implications of kidney injury • Guide to the NICU[a] and kidney care • How to advocate for your child • Clinic visit guide • Laboratory testing guide • Questions to ask clinician • Help with blood work and how to collect urine • Gamify education • Use videos • Intentionally build clinic or follow-up retention	Make caregivers feel guilty about their child's kidney disease risk
Enhances communication	—[b]	• Communication between patient and clinician • Questions and answers space, or frequently asked questions • NICU doctor livestream video • Communication between clinicians • Share laboratory results	Avoid increasing work burden of clinicians
Make scheduling appointments easier	—	• Scheduling • Help stacking and coordinating appointments to one visit • Appointment tracking and reminders	—
Track and sense make of laboratory results for caregivers	• Explain and interpret laboratory results	• Longitudinal tracking of laboratory results • Alert for concerning laboratory results	—
Identify treating health care team for caregivers	—	• Staff profiles and list	—
Support for caregivers	—	• Offer community and support	—
Help caregivers	—	• Note taking, keeping resources together • Longitudinal life of tool • Custom to patient	—
Access information outside of the stressful NICU setting	• App • Website • Printed materials	• Translate into different languages • Content on tablet at hospital	—

[a]NICU: neonatal intensive care unit.

[b]N/A: not applicable.

The research team then reviewed each item on the continuum and made decisions about what should be included in the final tool. The research team also discussed which of the items from the middle 2 sections should be included. The research team reviewed the bonus features and decided which of these to include in the final tool. Decisions were made based on how well the item would address the original objective.

Prototypes and Feedback

The research team created 2 prototype web pages to illustrate what a final tool might look like (Multimedia Appendix 3). For example, the home page included information about poor kidney development, potential kidney injury in the NICU, and how this may lead to the need for long-term kidney monitoring. It also contained a still from a video that might exist where a clinician explains NICU kidney injuries. The home page acts as the basic information for caregivers, while the rest of the site offers additional details. The menu items included: "about kidney monitoring," "common kidney tests," "talking with your child's doctor," and "caregiver support." Caregivers and clinicians reviewing the prototypes were supportive of the categories of information and content provided. They also appreciated the overall design of the prototype webpage. In general, they wanted caregiver stories with diverse people and languages, as well as more detailed information and research.

Discussion

We conducted a qualitative study examining caregiver attitudes and preferences toward the communication of kidney health by clinicians in the NICU setting. Our results suggest opportunities for improving communication about the risk of long-term kidney disease between caregivers and clinicians. Caregivers' primary concerns were the type and depth of information conveyed and

the time at which it was communicated. Both caregivers and clinicians emphasized the importance of collaboration between the NICU team and the primary care provider to ensure they were on the same page about the necessity of kidney monitoring.

This study represents the first attempt, to our knowledge, to develop a set of clear approaches to communicating kidney health and the risk of CKD in the NICU. Our findings are in concert with a recently published survey of caregivers with infants diagnosed with necrotizing enterocolitis during their NICU admission [17]. Both studies found that caregivers desired accurate and timely information to inform care and improve communication. Furthermore, other studies suggest that information gathering is an important coping mechanism for stress while their child is in the NICU [31]. Education-based programs have additional benefits for caregivers, including improved parental mental health outcomes, stronger beliefs in their parental role, and increased parental engagement [32]. The timely and family-centered provision of information and education is an essential aspect of family-centered care, which has increased parent engagement and satisfaction as it has become more widely used in NICUs over the last decade [33].

One challenge in neonatal kidney health clinical care and research studies is the low rate of kidney-specific follow-up for infants [34,35]. Studies suggest that, while multifactorial, contributing factors include poor provider and caregiver awareness of the risk of long-term kidney disease, a lack of family communication, and a perceived inability to change the course of disease with care [36]. Furthermore, siloing of care and electronic health care records which do not follow patients from health encounter to health encounter limit the ability of caregivers and clinicians to carry health information with them throughout the medical system. The development of improved communication with caregivers during and after their NICU stay is paramount to improving not only clinical care but also research studies of long-term kidney health, which are often stymied by poor retention. Our approach to kidney health communication was developed not by expert consensus of clinicians, as is often the case in similar studies, but by directly engaging with caregivers who have had infants admitted to the NICU who are at risk of long-term kidney disease. We believe this will result in a far more effective communication strategy that is more acceptable to families and increases the efficacy of subsequent follow-up.

There are several important limitations to this study. First, owing to the relatively small sample size and narrowness of the study population (eg, caregivers of infants at risk for CKD in the NICU), it is difficult to ascertain the broad generalizability of these findings. However, we attempted to recruit caregivers of various ages and backgrounds, at varying time periods out from their child's NICU stay (eg, 6 months post-NICU discharge vs 2 years post-NICU discharge) in order to improve generalizability to our larger population. Second, the design methods used are novel in health-related research, but they have been well-established in service and product design. Finally, the subjects we recruited were a convenience sample of nonconsecutive caregivers seen at our pediatric nephrology clinic who were willing to participate in research and may not accurately represent a random sample of our patient population.

Despite these limitations, this study represents an important first step in improving communication about kidney health to caregivers and families of those at high risk of kidney disease. The next step in this project is to further develop this communication tool based on caregiver and clinician guidance and to implement the tool in the NICU. Based on the above results and guidance from participants in this project, we are developing a website for family-centered kidney health information and plan to continue to gather input from caregivers to better understand the best ways to present and organize information, how to provide real-world experience and perspectives, and what information caregivers want at specific times during and after their child's NICU admission. Caregivers of infants admitted to the NICU will be given access to the revised communication tool developed in this study. We will then further assess the impact of the communication tool on their understanding of kidney health, the risk of long-term kidney disease, and follow-up patterns.

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Conflicts of Interest

None declared.

Multimedia Appendix 1
Process of human-centered design process to create family-centered communication tool.
[PNG File , 364 KB - jopm_v15i1e45316_app1.png]

Multimedia Appendix 2
Parent comment on barriers and drivers of kidney monitoring and communication strategies.
[DOCX File , 15 KB - jopm_v15i1e45316_app2.docx]

Multimedia Appendix 3
Preliminary prototypes developed.
[PNG File , 495 KB - jopm_v15i1e45316_app3.png]

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Abbreviations

AKI: acute kidney injury
CKD: chronic kidney disease
NICU: neonatal intensive care unit

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Original Paper

Acceptability of Automated Robotic Clinical Breast Examination: Survey Study

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Abstract

Background: In the United Kingdom, women aged 50 to 70 years are invited to undergo mammography. However, 10% of invasive breast cancers occur in women aged ≤45 years, representing an unmet need for young women. Identifying a suitable screening modality for this population is challenging; mammography is insufficiently sensitive, whereas alternative diagnostic methods are invasive or costly. Robotic clinical breast examination (R-CBE)—using soft robotic technology and machine learning for fully automated clinical breast examination—is a theoretically promising screening modality with early prototypes under development. Understanding the perspectives of potential users and partnering with patients in the design process from the outset is essential for ensuring the patient-centered design and implementation of this technology.

Objective: This study investigated the attitudes and perspectives of women regarding the use of soft robotics and intelligent systems in breast cancer screening. It aimed to determine whether such technology is theoretically acceptable to potential users and identify aspects of the technology and implementation system that are priorities for patients, allowing these to be integrated into technology design.

Methods: This study used a mixed methods design. We conducted a 30-minute web-based survey with 155 women in the United Kingdom. The survey comprised an overview of the proposed concept followed by 5 open-ended questions and 17 closed questions. Respondents were recruited through a web-based survey linked to the Cancer Research United Kingdom patient involvement opportunities web page and distributed through research networks' mailing lists. Qualitative data generated via the open-ended questions were analyzed using thematic analysis. Quantitative data were analyzed using 2-sample Kolmogorov-Smirnov tests, 1-tailed *t* tests, and Pearson coefficients.

Results: Most respondents (143/155, 92.3%) indicated that they would definitely or probably use R-CBE, with 82.6% (128/155) willing to be examined for up to 15 minutes. The most popular location for R-CBE was at a primary care setting, whereas the most accepted method for receiving the results was an on-screen display (with an option to print information) immediately after the examination. Thematic analysis of free-text responses identified the following 7 themes: women perceive that R-CBE has the potential to address limitations in current screening services; R-CBE may facilitate increased user choice and autonomy; ethical motivations for supporting R-CBE development; accuracy (and users' perceptions of accuracy) is essential; results management with clear communication is a priority for users; device usability is important; and integration with health services is key.

Conclusions: There is a high potential for the acceptance of R-CBE in its target user group and a high concordance between user expectations and technological feasibility. Early patient participation in the design process allowed the authors to identify key development priorities for ensuring that this new technology meets the needs of users. Ongoing patient and public involvement at each development stage is essential.

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KEYWORDS

breast cancer detection; automated diagnosis; breast examination; health care robotics; patient and public involvement; participatory design; user acceptability; mammography; breast cancer

Introduction

Background

Breast cancer is the leading cause of cancer mortality in women worldwide [1]. Almost 11,400 women a year died of breast cancer in the United Kingdom between 2015 and 2017 [2]. However, the mortality rate of breast cancer is falling, with a reduction from 60 per 100,000 in 1989 to 33 per 100,000 in 2017 [2,3]. This trend correlates with the introduction of widespread breast cancer screening using x-ray mammography [4]. In the United Kingdom, mammography is offered to women aged 50 to 70 years through the National Health Service (NHS) Breast Cancer Screening Programme [3]. Screening is estimated to reduce the relative risk of breast cancer mortality by 20% [5] and is linked to many lives saved each year [6].

However, mammography is not suitable for all groups who could benefit from breast cancer screening [7]. For example, 10% of invasive breast cancers occur in younger women (aged <45 years) in the United Kingdom at a rate of 235 per 100,000 [8], a group for whom mammography is not recommended because of its considerably decreased sensitivity in dense breast tissue [7]. This is particularly concerning as young women diagnosed with breast cancer are at higher risk of developing aggressive subtypes and have a poorer prognosis [9]. Mammography is also inappropriate for pregnant women as the low-dose radiation used poses a potential risk during lactation [9]. Furthermore, some women may be unable to tolerate mammography because of pain or discomfort [10]. A breast cancer screening modality that extends services to these groups has the potential to save years of life [11]. Identifying a screening alternative has been challenging. The most effective means of diagnosis (eg, triple assessment and magnetic resonance imaging) are often invasive or costly and unfeasible as screening modalities [12].

A promising alternative is clinical breast examination (CBE) [13,14]. A recent randomized controlled trial of CBE breast cancer screening involving >150,000 women in India demonstrated a 15% reduction in breast cancer–related mortality and a 10% relative risk reduction in the diagnosis of stage-III or stage-IV disease [15]. An overview of systematic reviews assessing the effectiveness of CBE screening identified indirect evidence that CBE has the same effect as mammography when performed well [16]. However, there are several challenges to ensuring that CBE is consistently "well performed." CBE screening effectiveness may be affected by variations in examination proficiency, training of health care professionals (HCPs), and a lack of standard documentation [17-21].

Recent advances in technology may provide a solution to these challenges. Robotics-assisted procedures have expanded rapidly in recent decades [22,23], and existing literature suggests that health users are increasingly more accepting of artificial intelligence (AI) and machine learning algorithms in cancer screening [24-26]. It is theoretically feasible to create a fully automated robotic CBE (R-CBE) platform by combining soft robotic technology and machine learning algorithms trained by breast specialists. This could offer much-needed standardization of CBE. R-CBE also has the potential to extend screening services to currently underserved groups as it is not reliant on radiation or affected considerably by tissue density. As health policy makers are discussing a risk-stratification approach to breast cancer screening, the cheap and low-risk modality of R-CBE may find further use as part of a strategy to classify the personalized risk level of an individual by measuring physiological properties such as mammographic density [27,28].

The Automated Robotic Examination Intelligent System (ARTEMIS), a novel robotic system for automated CBE, is currently being developed by our research team with support from Cancer Research United Kingdom (CRUK). ARTEMIS aims to combine soft robotic technology with a machine learning platform to allow for fully automated CBE and interpretation of results. The platform could be used by women without direct clinical supervision (Figure 1). A prototype is currently in the early stages of development [29-31]. Although such a platform may be capable of effectively performing and interpreting CBE, the voices of potential users are essential in determining how this should be designed and implemented. Creating technology and a service that is acceptable to end users (and meets their needs) will be crucial in determining the uptake of this type of technology.

Very little published literature is available on the acceptability of intelligent systems that interface directly and independently with users. We identified only 1 study assessing the acceptability of autonomous robotic systems that interface directly with users in health care. This study used robotics to perform basic patient assessment tasks (eg, measuring vital signs and inserting intravenous catheters) and concluded that this would be acceptable [32]. We did not identify any publications exploring the acceptability of intelligent robotic services that directly interact with users in cancer screening or diagnostics.

Figure 1. Automated Robotic Examination Intelligent System user interface (conceptual diagram).

Objectives

This study investigated whether R-CBE is theoretically acceptable to potential users and explored the attitudes, perspectives, and concerns of women regarding the use of intelligent robotic technology in breast cancer screening. It identified key factors that determine whether (and how) the technology would meet the needs of patients, allowing these to be integrated into the prototype design. We adopted the definition of acceptability proposed by Sekhon et al [33]: "a multi-faceted construct that reflects the extent to which people receiving a healthcare intervention consider it to be appropriate, based on anticipated or experienced cognitive and emotional responses to the intervention." We conducted a web-based survey of 155 women in the United Kingdom to investigate the following questions: (1) Is there a perceived need for R-CBE? (2) What elements of the R-CBE user interface are most important to women? (3) Is this technology likely to be acceptable to potential users? To the best of our knowledge, this is the first study assessing the acceptability of a fully automated and intelligent patient examination system that interacts directly with users for breast cancer screening.

Methods

A mixed methods approach was used to collect and analyze qualitative and quantitative data from the survey.

Survey Development

The survey consisted of 5 open-ended questions and 17 closed questions with a separate free-text section for respondents to share additional information. A brief overview of the proposed ARTEMIS concept was provided to respondents (Multimedia Appendix 1). This included Figure 1 and a description of how the user might interact with the palpation platform but had no technical details or any information on the accuracy of the device. Our aim was to allow the respondents to freely think about factors that might affect their use of the hypothetical service without imposing any of our priors.

Key constructs were identified based on a review of the health intervention acceptability and health technology literature, our broader knowledge of health technology, and support from the CRUK patient and public involvement specialist team and the London In Vitro Diagnostics Co-operative. We did not identify a fully validated model suitable for our research questions; instead, 2 frameworks were combined with questions selected to cover essential constructs from both. The first was the Theoretical Framework of Acceptability for health care interventions proposed by Sekhon et al [33]. The second was the Unified Theory of Acceptance and Use of Technology developed by Venkatesh et al [34], which has been widely used in research exploring the acceptance of ITs [35]. The resultant key constructs encompassed affective attitudes, perceived effectiveness, ethicality, self-efficacy, effort expectancy, social influence, and facilitating conditions.

The questions were carefully designed to illuminate implicit assumptions and ensure that all key constructs were considered while maintaining an accessible and nonleading language [36]. After a multistage drafting process, the survey was collated and tested on close contacts and members of the associated research department for appropriateness, readability, and ease of use to produce a final draft (Multimedia Appendix 1).

Closed questions allowed us to quantify the overall level of acceptability and desirability of specific features of the service (eg, interface, timing, and preferred location). Thematic analysis of qualitative data provided insights into the quantitative findings. This added richness to our understanding of potential users' perspectives and attitudes toward the proposed ARTEMIS R-CBE and allowed us to build a more complete picture of acceptability.

Recruitment and Data Collection

Female respondents aged between 20 and 70 years were recruited through a web-based survey linked to the CRUK patient involvement opportunities web page and newsletter and the People in Health West of England and Imperial Human Behaviour and Experience network mailing lists. This nonprobability, voluntary response sampling strategy was chosen because of its quick recruitment rate and ability to serve the exploratory nature of the study. With no hypothesis to test, the aim of the survey was to develop an initial understanding of the needs of the population, and so the bias introduced by self-selection was considered acceptable.

The 15-minute web-based survey was hosted on Qualtrics (Qualtrics International Inc), and 2 attention-check questions were added to ensure that respondents read each question carefully and also to exclude nonhuman (automated) respondents; this resulted in the expulsion of 1 set of responses because the attention questions were answered incorrectly. A further 15 questionnaires were discarded because they were incomplete, including incomplete attention questions, and 3 were discarded because they did not meet the inclusion criteria. This meant that, of 174 responses initiated, 155 (89.1%) completed the survey over 6 weeks between August 2020 and September 2020. Summing the size of each of the mailing lists gives a response rate of 9.26% (155/1674).

Data Analysis

Quantitative analysis was conducted in MATLAB (MathWorks), and differences between groups based on demographics were identified using a 2-sample Kolmogorov-Smirnov test, explored using 1-tailed *t* tests, and reported where significant (full results available in the data set referenced in the Data Availability section). Pearson correlations were calculated where appropriate to quantify the strength of the associations. CIs were calculated for ranked questions assuming that the preferences were equidistant (1>2>3...).

Qualitative data were analyzed using a method designed around thematic analysis [36]. This allows for detailed exploration of patterns across a data set using a latent approach, with researchers gaining a rich understanding of respondents' perspectives [36].

Themes were identified after familiarization with the open-text responses. To this end, 2 researchers independently identified a set of key themes within the responses, chosen with relevance to identifying the factors that influenced the respondents' acceptance of the hypothetical technology. After combining these sets of themes, a single researcher divided each theme into concepts that tightly grouped responses within each theme. Salient ideas from these grouped concepts were then extracted to describe the outcomes of the responses as a whole.

The raw data are available from the source provided in the Data Availability section at the end of this paper.

Ethics Approval

The study received ethics approval from the Imperial College Research Ethics Committee (20IC6129).

Results

Quantitative Results

Demographics

The average age of the respondents was 49.8 (SD 12.7; range 21-70) years. "White" ethnic (142/155, 91.6%) and university-educated (119/155, 76.8%) backgrounds were overrepresented among survey respondents. Our study population also had a higher incidence of personal history of breast cancer (28/155, 18.1%) compared with the general adult population (4.46% [8]). Respondents were overwhelmingly in favor of screening programs (143/155, 92.3%) and the use of technology in health care (146/155, 94.2%). The demographic data are summarized in Table 1, and attitudes toward screening and technology in health care in general are summarized in Table 2.

Table 1. Respondent demographics (N=155).

Demographics	Values, n (%)
Age (years)	
21-44	54 (34.8)
45-59	48 (31)
60-70	51 (32.9)
Did not complete	2 (1.3)
Ethnicity	
White British	121 (78.1)
Other White	21 (13.5)
Black African	4 (2.6)
Indian	1 (0.6)
White and Black African	1 (0.6)
Pakistani	1 (0.6)
White and Black Caribbean	1 (0.6)
Chinese	1 (0.6)
Prefer not to say	3 (1.9)
Highest qualification	
Bachelor's degree or higher	119 (76.8)
Vocational qualification (ONC[a], BTEC[b], or NVQ[c])	13 (8.4)
A-Levels (or equivalent)	13 (8.4)
GCSE[d] or O-Levels (or equivalent)	10 (6.5)
History of diagnosis of cancer	
Any (including breast cancer)	42 (27.1)
Breast cancer	28 (18.1)

[a]ONC: Ordinary National Certificate.

[b]BTEC: Business and Technology Education Council qualification.

[c]NVQ: National Vocational Qualifications.

[d]GCSE: General Certificate of Secondary Education.

Table 2. Respondents' attitudes toward breast cancer screening and technology in health care (N=155).

Questions and responses	Values, n (%)
Do you think routine cancer screening tests are a good idea?	
Yes	143 (92.3)
No	4 (2.6)
Don't know	8 (5.2)
What do you think of increased use of new technology in health care?	
Very bad idea	0 (0)
Bad idea	5 (3.2)
Good idea	48 (31)
Very good idea	98 (63.2)
Don't know	5 (3.2)

Overall Opinion Toward the Device

Provided the R-CBE was as good as an HCP, 92.3% (143/155) of respondents said that they would either "definitely" (104/155, 67.1%) or "probably" (39/155, 25.2%) use an R-CBE service if it were offered. In comparison, 89.7% (139/155) of respondents said that they would "definitely" (92/155, 59.4%) or "probably" (47/155, 30.3%) use a service offering CBE by a trained HCP (Figure 2). This indicates that the answers to the 2 questions were similar, with a slight preference for R-CBE (2-sample Kolmogorov-Smirnov test; P=.40). Willingness to use an R-CBE service was moderately correlated with respondents' likelihood of using new technology in general (r_{155}=0.4014; P<.001).

Respondents were asked to indicate which factors would make them more likely to use R-CBE. The most popular option was receiving a "faster referral to specialist breast services" if required (144/155, 92.9% of respondents selected this option) and being able to drop in and use the device without an appointment (108/155, 69.7% of respondents). Other factors that influenced anticipated use are shown in Table 3.

Figure 2. Overall opinion of the device. This demonstrates that the idea of a robotic system appeals to some respondents more so than the status quo. CBE: clinical breast examination.

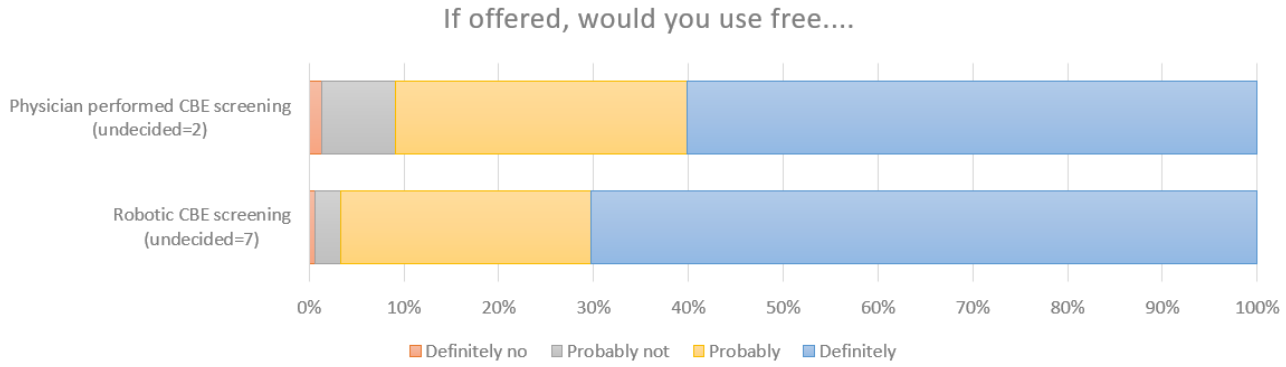


Table 3. Factors to improve uptake, which provides insights into the respondents' understanding of how a robotic system might best be of benefit to them (N=155).

What would make you more likely to use R-CBE[a]	Values, n (%)
Faster referral to a specialist	93 (60)
Drop-in appointments	70 (45.2)
Knowing what to expect before the appointment	68 (43.9)
My GP[b] seeing the results	61 (39.4)
Confidential results	59 (38.1)
More technical information	39 (25.2)
Information on data protection	36 (23.2)

[a]R-CBE: robotic clinical breast examination.

[b]GP: general practitioner.

Device Features

The respondents favored the use of soft (rather than hard) robotic parts for the aspects of the device that would be in contact with their skin. Device features considered to be of most importance were availability of information on access to support from an HCP, appointment availability, cleanliness, and regular updates on examination progress throughout the procedure. The results are presented in Table 4.

A comparative analysis of the age groups revealed 3 significant differences. Each respondent scored a selection of features on a scale of 1 to 5. The age group of >60 years (the oldest) considered ease of appointment availability to be less important compared with the 2 younger age groups (>60 years vs 45 to 59 years: *mean difference [MD]*=0.37 and P=.02; >60 years vs <45 years: *MD*=0.43 and P=.04). Conversely, the age group of <45 years considered it less important to be able to adjust the speed of the device (<45 years vs 45 to 59 years: *MD*=0.59 and P=.04; <45 years vs >60 years: *MD*=0.87 and P=.001) or for the device to have disposable parts (<45 years vs 45 to 59 years: *MD*=0.89 and P=.002; <45 years vs >60 years: *MD*=0.70 and P=.02).

Table 4. Relative importance of device features. "On a scale from 1 (not at all important) to 5 (essential), how important is it that..."

Feature	Score, mean (95% CI)
The device provides links to support from HCPs[a]	4.26 (4.12-4.39)
Appointments are easily available	4.16 (4.04-4.28)
Information about the cleaning of the booth is available	4.10 (3.96-4.25)
The examination provides constant updates	4.08 (3.96-4.21)
The device is close to home or work	3.63 (3.49-3.78)
Parts of the device that are in contact with the skin are disposable	3.34 (3.14-3.55)
I am able to adjust the speed of the device's parts that are in contact with the skin	2.76 (2.58-2.94)

[a]HCP: health care professional.

Location

Most respondents (130/155, 83.9%) preferred the booth to be located at a site associated with health care. The most popular location was at a general practitioner surgery, which generally provides point-of-contact care and triage between patients and specialist health services in the United Kingdom, followed by "inside a pharmacy." Options not associated with health care (such as at a shopping center or in the workplace) were less popular. This difference was statistically significant. Location preference is shown in Table 5. The age group of >60 years favored the shopping center more compared with the other age groups (>60 years vs 45 to 59 years: *MD*=0.50 and *P*=.02; >60 years vs <45 years: *MD*=0.57 and *P*=.008).

Table 5. Location preference.

Rank	Option	Rank, mean (95% CI)
1	GP[a] surgery	1.41 (1.23-1.50)
2	Pharmacy	2.15 (1.97-2.20)
3	Shopping center	3.77 (3.47-3.82)
4	Work	3.84 (3.52-3.91)

[a]GP: general practitioner.

Length of Examination

Most respondents (153/155, 98.7%) were willing to be examined for up to 10 minutes, 82.6% (128/155) were willing to be examined for up to 15 minutes, and 56.8% (88/155) were willing to be examined for 20 minutes. Interestingly, only 22.6% (35/155) of the respondents considered the time taken to carry out the examination to be "Quite important" (34/155, 21.9%) or "Essential" (1/155, 0.6%). When asked to rate "how important is it that the examination does not take longer [than the duration the respondent indicated]" on a scale of 0 (not at all important) to 5 (essential), the mean rating was 1.46. However, respondents who preferred a shorter examination duration were statistically more likely to report that it was important that the examination last no longer than they had indicated (r_{153}=0.53; *P*<.001). These results are shown in Figures 3 and 4.

Figure 3. Cumulative tolerable examination duration. Nearly all respondents (153/155, 98.7%) were happy with an examination lasting up to 10 minutes, with a substantial minority (88/155, 56.8%) happy with a duration of up to 20 minutes.

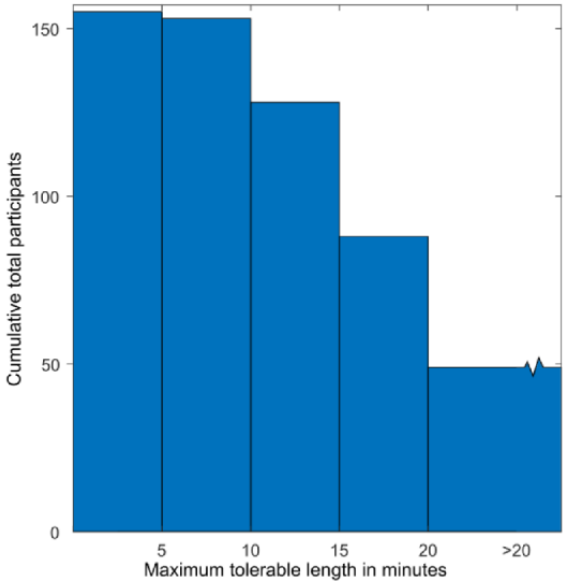
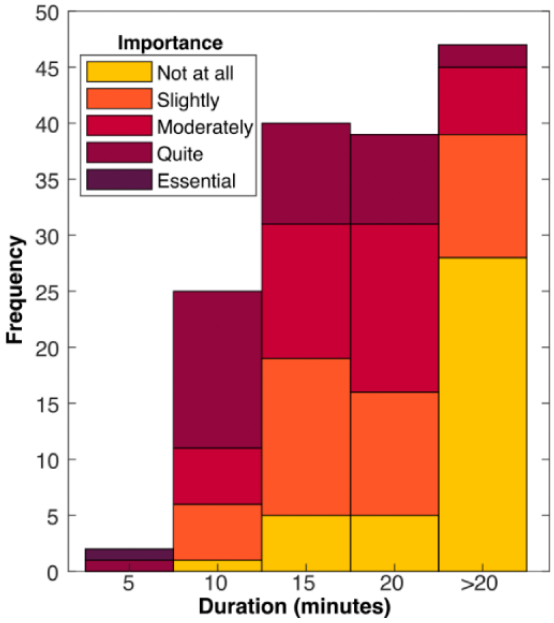


Figure 4. Tolerable length of examination. The respondents who preferred a shorter examination also considered duration a more important factor.



Communication of Results

Most users (128/155, 82.6%) preferred to receive information directly from the device, either displayed on the screen with a printout (mean rank 2.26) or received via email (mean rank 2.47). These options were statistically significantly more popular than the results being emailed to their physician first. This was true both in the case of a *normal* (mean rank 5.02) and an *abnormal* (mean rank 3.72) result. In the event of an *abnormal* result, the option "email to my doctor first" increased in preference (from sixth to fourth in the average rank) but remained comparatively unpopular. The most popular option for results communication was through a combination of written information and pictures (mean rank 1.62). Respondents without a university diploma or equivalent ranked seeing their results on-screen without a printout significantly higher than those with a university diploma or equivalent (healthy: *MD*=0.57 and *P*=.08; *abnormal*: *MD*=0.89 and *P*=.02). This suggests that the level of education may be an important discriminant when considering how results are communicated. Respondents highly valued the inclusion of information on appropriate follow-up and alternative explanations for identified *abnormalities*. The results are summarized in Tables 6-8.

It is worth noting that 6.5% (10/155) of respondents used the open-text "other" option to indicate that they would want to receive results from an HCP and not from the R-CBE device itself. All respondents (155/155, 100%) ranked this as their number 1 preference.

Table 6. Information receipt preferences.

Option	No referral advised, mean rank (95% CI)	Referral advised, mean rank (95% CI)	Rank change
Immediately on-screen+printout	2.26 (2.02-2.50)	2.41 (2.20-2.62)	−0.15
Emailed later	2.47 (2.26-2.68)	2.66 (2.49-2.83)	−0.19
Immediately on-screen	2.76 (2.57-2.96)	3.30 (3.06-3.53)	−0.52
SMS text message	4.10 (3.90-4.29)	4.61 (4.37-4.75)	−0.46
Posted later	4.24 (4.07-4.41)	4.25 (4.05-4.44)	−0.01
Emailed to physician first	5.02 (4.82-5.21)	3.72 (3.43-4.00)	+1.31
Other	7.40 (7.11-7.68)	6.40 (6.16-6.63)	0

Table 7. Information display preferences.

Rank	Option	Rank, mean (95% CI)
1	Written and pictures	1.62 (1.48-1.76)
2	Interactive app	2.02 (1.84-2.20)
3	Written only	2.53 (2.38-2.68)
4	Verbal summary	3.785 (3.6-3.97)

Table 8. Information content preferences (N=155).

What information would you like included in your results	Respondents, n (%)
Details on follow-up when referral is recommended	153 (98.7)
How to book a future R-CBE[a] appointment	151 (97.4)
Other causes for an “abnormal” finding	122 (78.7)
Links to emotional support	108 (69.7)

[a]R-CBE: robotic clinical breast examination.

Qualitative Results

Overview

Qualitative analysis of the free-text responses identified the following seven superordinate themes with respect to R-CBE: (1) women perceived that R-CBE has the potential to address limitations in current screening services, (2) R-CBE may facilitate increased user choice and autonomy, (3) ethical motivations for supporting R-CBE development, (4) accuracy (and users’ perceptions of accuracy) is a priority, (5) results management with clear communication is a priority for users, (6) integration with health services is key, and (7) device usability is important. These themes are summarized in the following sections. Quotes from the respondents illustrating the themes are shown in Table 9.

Table 9. Themes from thematic analysis with supporting quotes.

Theme and concept	Quotes
R-CBE[a] has the potential to address limitations in current services	
Provides reassurance	• "I worry about my breast health. It would be reassuring to be able to check for irregularities." [Respondent 090] • "...to be able to regularly monitor for something like breast cancer would give me peace of mind." [Respondent 008]
Reluctance to "waste" physicians' time	• "I find [breast examination] difficult to do myself and don't like to take up doctors time very often." [Respondent 032] • "I would also like regular check-ups and understand GPs need to prioritise other appointments." [Respondent 097]
Negative experiences with mammography	• "When I have a mammogram it really hurts me. I often say that the machine is like torture." [Respondent 129] • "I would welcome any solution that is pain free." [Respondent 129]
Embarrassment during clinical examination	• "Every time I see breast screening on TV there is a picture of women with a completely naked top half. This makes me feel very uncomfortable and puts me off screening." [Respondent 053] • "I would rather have a machine examine my breasts than a doctor. It would eliminate the feeling of embarrassment." [Respondent 036] • "...lack of human contact may encourage more women to use it." [Respondent 087]
Anxiety associated with awaiting results	• "Every time I have a mammogram, I panic for 2-3 weeks waiting for the results." [Respondent 134] • "If results are available immediately then that's better than waiting for test results and stops stress and anxiety." [Respondent 106] • "The possibility of having instant results is amazing." [Respondent 142]
R-CBE may facilitate increased user choice and autonomy	
Choice over appointment time, frequency, and location	• "...it's is [sic] more convenient if you have a bigger choice over appointment times." [Respondent 039] • "Freedom to choose when to use the device." [Respondent 029] • "...hopefully accessibility (location/appointments) are easier than going to the GP." [Respondent 049] • "...this would allow more frequent checks." [Respondent 147]
Increased sense of autonomy	• "...the opportunity to be firmly in control of ones [sic] own health concerns is appealing." [Respondent 143] • "I believe the autonomy of this device may encourage more people to come forth for screening." [Respondent 132]
Ethical motivations for supporting R-CBE	
Support for population screening in general	• "I like to spread the word about health screening, it's very important to look after your health." [Respondent 053] • "I would support anything that encourages people to be tested." [Respondent 054]
Potential to increase access for underserved populations	• "I think it is very important that women can have regular breast examinations that start at a younger age that [sic] mammograms!" [Respondent 007] • "Digital Automation seems to be one way of improving life chances for black Cancer patients like myself." [Respondent 144] • "[a family member] has a learning difficulty, is deaf and is a wheelchair user. It has not been possible for her to have the benefit of regular breast screening. I am hopeful that this new device will help women like her in the future." [Respondent 143]
Reduced burden on the NHS[b]	• "Technology is advancing and the population is growing. Using this technology in health care will help to free up our medical staff so that they can use their much needed skills working it [sic] areas where only human intervention is possible." [Respondent 063] • "It would seem to be an efficient screening tool that allows precious medically trained staff to do other jobs a machine cannot do." [Respondent 111]
Accuracy, and users' perception of accuracy, is a priority	
R-CBE is only acceptable if users are convinced of its accuracy	• "I think it is a very good idea...provided there is a definite level of assured accuracy." [Respondent 062] • "I would use it if I had confirmation that results are accurate." [Respondent 054]

Theme and concept	Quotes
Factors that increase confidence in accuracy	• "...if the technology and device is proven through appropriate clinical studies." [Respondent 155] • "...how often it gets the diagnosis right, how often it gets it wrong." [Respondent 022] • "MHRA [Medicines and Health care products Regulatory Agency] approval." [Respondent 041] • "...some sort of checking procedure e.g. [sic] every 50th person is called in for manual checks." [Respondent 054]
Suitable results management with sensible communication	
Sensitivity concern about receiving results from R-CBE	• "I worry about the emotional impact of an abnormal result being given via automated means." [Respondent 006] • "I think a human can usually be gentler with the feelings of patients." [Respondent 022]
Rapid results are preferred	• "[I] wouldn't want doctor involvement to delay my getting the result." [Respondent 042] • "I would much prefer the results at the time of the test." [Respondent 134]
Factors that optimize user experience when receiving results	• "I'd want to know more about what an 'abnormal' result might mean—does it definitely mean cancer, or could it mean something else?" [Respondent 100] • "If there is an abnormal result it will cause an amount of worry and anxiety and so any additional information that can be provided alongside the results such as emotional support and links to further information would be really useful." [Respondent 142] • "[if] the results are abnormal...an automatic urgent appointment should be made by the GP straight away." [Respondent 079]
Confidentiality and privacy are essential	• "...all the physical privacy and data privacy issues [need to be] well thought through." [Respondent 059] • "To be screened in a booth, it would have to be entirely 100% privacy proof, confidential, and safe." [Respondent 134]
Integration with health services is key	
High trust placed in the NHS	• "I would use a machine if it ran in tandem with NHS services." [Respondent 022] • "If it's recommended by my GP or other relevant HCP." [Respondent 141] • "I would expect it to complement other services not replace them." [Respondent 145]
Geographic proximity to other health services	• "I think I would feel more comfortable if the service was in a health care setting (e.g. GP/pharmacy), rather than in a more public space (e.g. work)." [Respondent 014] • "I think the location should be somewhere linked to medical care/support—even if just near a first aider's office." [Respondent 146]
Device usability is important	
Clear instructions required	• "...if the instructions are fool-proof I think I could manage it." [Respondent 063] • "[needs] clear and understandable for everyone." [Respondent 082] • "A video demo would be helpful to maybe watch before attending." [Respondent 148]
Clear plan for managing technical difficulties	• "I might need a little reassurance that the machine wasn't going to run amok." [Respondent 149] • "My only reservation was if it went wrong and either used the wrong pressure or wouldn't unclamp from the breast." [Respondent 055] • "Where to get help if the device didn't work or stopped working during examination." [Respondent 145] • "...a panic or immediate stop function [with] the ability to cancel and walk away." [Respondent 151]

[a]R-CBE: robotic clinical breast examination.

[b]NHS: National Health Service.

Women Perceive That R-CBE Has the Potential to Address Limitations in Current Screening Services

The limitations of current breast cancer screening services were raised frequently, and respondents perceived that R-CBE has the potential to address some of these limitations. "Check-ups" could provide regular reassurance lacking in current services. Respondents recognized that they could regularly self-examine (but lacked confidence to do so) or request regular examinations from a health practitioner (but did not want to waste the physician's time). Pain associated with mammography was the most frequently cited limitation of breast cancer screening. Many respondents assumed that soft robotics would be more comfortable than a mammogram. R-CBE could also reduce the embarrassment of being seen unclothed by an HCP during mammography or CBE. Some respondents believed that a fully automated service that reduced this embarrassment was preferable to direct human involvement. Long waiting times to

receive screening results were associated with anxiety, and the possibility of receiving rapid results from automated technology was highly appealing. This theme reflects the potential of R-CBE to address limitations in current services.

R-CBE May Facilitate Increased User Choice and Autonomy

R-CBE may be "more convenient" than other screening services, offering a wider choice of appointment times, location, and the frequency with which the service could be accessed. This increased choice over where and when, combined with the opportunity to complete screening without input from an HCP, was appealing and provided a sense of autonomy and control.

Ethical Motivations for Supporting R-CBE Development

Some respondents viewed R-CBE favorably on an ethical basis. For example, respondents suggested R-CBE (with the potential to be a convenient and accessible service) could increase screening among traditionally underserved populations such as young women, ethnic minorities, or people with disabilities. There was a desire to extend screening and cancer prevention on a population basis, irrespective of the modality, and strong support for the NHS. Respondents indicated that they would accept R-CBE if it reduced the burden on the NHS and HCPs. This reflects an underlying assumption that an automated device screening service would reduce the burden on the NHS. This assumption is explored further in the Discussion section. This theme indicates support for the R-CBE concept based on the respondents' broader attitudes and ethical beliefs.

Accuracy, as well as Users' Perception of Accuracy, Is a Priority

Acceptance of R-CBE was conditional, and respondents identified several factors required for R-CBE to be trustworthy. Chief among these was accuracy. Unsurprisingly, the requirement that the device have high levels of accuracy was mentioned by most respondents (132/155, 85.2%) unprompted. There was no clear required accuracy threshold. Some respondents wanted to see a service that was "as good as a mammogram," others wanted to see a service "as good as a GP," and others still "would use the device on the condition that it was better than a doctor." However, there was a consensus that users should be provided with enough information to make their own informed decision as to whether R-CBE is accurate enough. Respondents suggested that users be given information on the sensitivity and specificity, ongoing monitoring of device performance, clinical trials completed, and regulatory approval to optimize trust. To be trustworthy, R-CBE must be highly accurate, and salient understandable information on how this accuracy is determined must be made available.

The Need for Suitable Results Management With Sensitive Communication

Communication of results in a sensitive manner was a key priority. Receiving screening results is anxiety-inducing, and the responses indicated that this is particularly true for technology-based services. Some respondents expressed concern about the ability of R-CBE to do this in a sufficiently sensitive manner. A small number of respondents felt that direct human involvement was essential in the event of an *abnormal* result. They felt strongly about this and described the idea of receiving an *abnormal* result from an automated device as "cold," "impersonal," and "abhorrent." However, more respondents reported that rapid availability of results outweighed this disadvantage. Options for optimizing direct R-CBE results delivery were identified. These included ensuring an efficient follow-up process, providing information on possible causes of an *abnormal* result (options other than malignancy), and providing guidance on where users could access support if needed. It was also important to respondents that results management be private and confidential and that detailed information on data storage be available.

This theme illustrates the need for efficient, sensitive, private, and secure processes for managing results that place users first. Providing sufficient information to service users may optimize the experience and minimize the anxiety associated with receiving results.

Integration With Health Services Is Key

Along with timely follow-up of *abnormal* results, functional integration with the health service was highly valued. Adequate integration with the health service appeared to increase user confidence in the new technology. A high degree of trust was placed in the NHS, and integration with this would lend credibility to R-CBE. It was important that the new technology be an adjunct to existing services without reducing access to general practitioners or current NHS services. Geographic proximity to existing health services was also viewed positively as respondents perceived that this could improve integration and access to support. The trustworthiness of R-CBE appears to depend not only on the device itself but also on the extent to which it is integrated into the existing health system.

Device Usability Is Important

Acceptability was conditional on R-CBE being easy to use. People must also be confident that they can use the device without compromising accuracy. The importance of clear instructions was highlighted; providing a short instructional video was a suggested method of ensuring this. There was also a degree of anxiety regarding the possibility of malfunctions. Respondents wanted a clear procedure for dealing with technical difficulties. Suggestions for this included an emergency stop function and a process for calling for assistance. Clear instructions, a plan for malfunctions, and an emergency stop button would provide peace of mind and respect women's autonomy by giving them control over the examination.

Discussion

Principal Findings

Responses were generally positive for a potential R-CBE service that is at least equivalent to a nonrobotic alternative. The overwhelming majority of respondents reported that they would use R-CBE screening if it were offered. Respondents recognized the potential of R-CBE to address an unmet need in current screening services by providing regular reassurance, reducing interpersonal embarrassment, reducing screening-associated pain, improving appointment availability, and offering rapid

results. All these are barriers to screening uptake recognized in the existing literature [37].

The survey showed that a high level of sensitivity and specificity of this technology is an essential factor for acceptability. User acceptance in our survey was dependent on R-CBE being a highly accurate system.

The results of this study also complement the existing literature on AI diagnostics, which suggests that the public has a high level of trust in computerized decision-making in health care and that AI in cancer screening is increasingly accepted [24-26].

The acceptance of R-CBE was qualified. Our results complement the existing literature [38] by identifying high levels of trust as an essential property for the uptake of robotic and automated systems. Our data identified factors that are necessary for an R-CBE service to be considered trustworthy. Key among these are accuracy, usability, and communication. Respondents' concerns regarding the lack of human connection, data privacy, and regulation of new health technology echoed similar concerns identified in a recent study exploring the public perception of AI mammography reading [26].

Our results indicate that most users are likely to accept autonomous screening if there is a well-established, efficient process for follow-up with a clinician if needed. This agrees with studies to date indicating that people are more accepting of intelligent systems working symbiotically with physicians or HCPs [24,39] but remain ambivalent about those that function independently [25].

This study provides important information to guide decision-making on R-CBE development, determine its viability as an investment, and inform our understanding of public attitudes toward intelligent health technology in cancer screening. Crucially, our results indicated a significant concordance between what is technically feasible and what is acceptable to users. For example, most respondents (128/155, 82.6%) were willing to be examined for up to 15 minutes and were also willing to receive results directly from ARTEMIS (in some format) rather than from an HCP. Research suggests that it is feasible to create an automated R-CBE service based on these acceptability characteristics [29-31].

Limitations and Future Directions

From these results, we believe that R-CBE may offer a more patient-focused option that has the potential to increase screening uptake provided it can perform examinations with sufficient sensitivity and specificity.

To develop technologies seeking to provide the service of R-CBE or similar, these results provide appropriate targets to be met when evaluating their expected acceptability. For example, several respondents supported R-CBE because it would reduce the burden on the NHS and free up time for HCPs. Although early detection and intervention could reduce progression to advanced disease (and, therefore, reduce the treatment burden on the NHS), this assumption is only valid if R-CBE detects early disease and allows for early intervention without overdiagnosis or excessive referrals to primary or specialist services.

As an investigative survey, the sample size was comparatively small, and the skewed distribution of demographic groups within the sample means that it was insufficiently powered to detect nuanced differences between them. A larger sample size with a demographic distribution representative of the wider population would be needed to identify whether the subtle differences in preference between demographic groups in this study are statistically significant and externally valid in the general population.

The demographics of the respondents were also not representative of the UK population. First, Black and minority ethnic groups were underrepresented. The data may not accurately capture the needs, thoughts, attitudes, and perspectives of these demographics. This is of particular concern as these groups are at an elevated risk of breast cancer and face the greatest barriers to screening [40,41]. Reaching these groups in future research is essential as they may benefit substantially from widening screening. Achieving this is likely to require targeted methods.

In addition, over three-quarters of our sample (119/155, 76.8%) had a degree-level education. Jonmarker et al [25] found a significant association between level of education and level of trust in technology. This is reflected in the very high levels of trust in technology reported in our sample. This reduces the generalizability of our results, with survey respondents being more likely to find R-CBE acceptable than the general population. The non–probability sampling used in this study may also introduce selection bias—it is possible that women who had a history of engaging with existing breast cancer screening programs were more likely to answer the survey, which might have contributed to overestimation of the acceptability of R-CBE screening. The particular method of electronic survey requires respondents to have ready access to a compatible device connected to the internet and be literate at using it, inherently excluding those who do not fulfill both criteria.

The ARTEMIS R-CBE is currently in development (part of this system is described in the study by Jenkinson et al [31]); the responses relate to a theoretical service. Further research will be needed to establish the acceptability of the specific service among users as development continues, as well as an assessment of its cost and accuracy. Future research would benefit from a larger and more diverse sample size that better represents the population. Our team is currently undertaking further qualitative research via focus groups to better understand the requirements of trustworthy and acceptable R-CBE and automated breast cancer screening more generally. Despite the limitations outlined previously, the survey data allowed us to identify key priorities among potential users and provide valuable information for the research team. These findings may provide insights for others working in automated health technology development, particularly for cancer screening.

Conclusions

R-CBE holds promise as a new modality of breast cancer screening. It could address limitations in current screening services, increase screening uptake, and provide a more patient-focused service. This investigative survey demonstrated

that there is potential for high levels of acceptability of R-CBE among its target user group and a high concordance between user expectations and technological feasibility. However, the acceptability of R-CBE is conditional on users being confident that it is accurate, easy to use, able to communicate results sensitively, and well integrated with health services. These findings will contribute directly to prototype development and will be of interest to other researchers developing automated cancer screening and related health technologies. This study highlights the fact that the development of new technologies raises ethical and practical issues. The importance of public and patient involvement in health technology development to address these issues should not be underestimated. Patient and public involvement at each stage of development will be key to ensure that any future service meets the needs of the public.

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Data Availability

The data sets generated for this study can be found on GitHub [42].

Conflicts of Interest

None declared.

Multimedia Appendix 1
Questionnaire.
[DOCX File , 203 KB - jopm_v15i1e42704_app1.docx]

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Abbreviations

AI: artificial intelligence
ARTEMIS: Automated Robotic Examination Intelligent System
CBE: clinical breast examination
CRUK: Cancer Research United Kingdom
HCP: health care professional
MD: mean difference
NHS: National Health Service
R-CBE: robotic clinical breast examination

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Original Paper

Older Adults' Experiences With Participation and eHealth in Care Coordination: Qualitative Interview Study in a Primary Care Setting

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Abstract

Background: Owing to the demographic changes in the elderly population worldwide, delivering coordinated care at home to multimorbid older adults is of great importance. Older adults living with multiple chronic conditions need information to manage and coordinate their care. eHealth can be effective for gaining sufficient information, communicating, and self-managing chronic conditions. However, incorporating older adults' health preferences and ensuring active involvement remain challenging. More knowledge is needed to ensure successful participation and eHealth use in care coordination.

Objective: This study aimed to explore multimorbid older adults' experiences with participation and eHealth in care coordination with general practitioners (GPs) and district nurses (DNs).

Methods: The study had a qualitative explorative approach. Data collection included semistructured interviews with 20 older adults with multimorbidity receiving primary care services from their GPs and DNs. The participants were included by their GPs or nurses at a local intermunicipal acute inpatient care unit. The data analysis was guided by systematic text condensation.

Results: We identified 2 categories: (1) older adults in charge of and using eHealth in care coordination, and (2) older adults with a loss of control in care coordination. The first category describes how communication with GPs and DNs can facilitate participation, the importance of managing own medication, and how eHealth can support older adults' information needs. The second category focuses on older adults who depend on guidance and help from their GPs and DNs to manage their health, describing how a lack of capacity and system support to be involved makes these adults lose control of their care coordination.

Conclusions: Being in charge of care coordination is important for older multimorbid adults. The results show that older adults are willing to use eHealth to be informed and to seek information, which ensures high levels of participation in care coordination. Future research should investigate how older adults can be involved in electronic information sharing with health care providers.

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KEYWORDS

care coordination; older adults; participation; eHealth; primary health care

Introduction

Older adults with multimorbidity (co-occurrence of two or more chronic conditions) [1] often experience challenges with care coordination and navigating the health care system [2-5]. Demographic changes in the elderly population worldwide have led to a necessity to treat and care for older adults with chronic conditions at home [6-8]. To meet these challenges, engaging older adults in managing their health and navigating health care services is essential and can reduce health care use [9-11]. Care coordination can be defined as the deliberate organization of patient care activities between two or more participants (including the patient) involved in a patient's care to facilitate the appropriate delivery of health care services. Organizing care involves the marshaling of personnel and other resources needed to carry out all required patient care activities and is often managed by the exchange of information among participants responsible for different aspects of care [3]. McDonald et al [3] emphasized that the patient's perspective is essential to care coordination. It is the patients who evaluate whether the coordination of care is experienced as sufficient [12]. The care coordination measurement framework describes different measures that illustrate what mechanisms meet the patient's needs and the delivery of high-quality health care services. Key care coordination mechanisms described by McDonald et al [3] are communication among involved personnel, information sharing, facilitating transition, supporting patients' self-management goals, and assessing patients' needs and goals. Other broader approaches to care coordination are medication management and health information technology–enabled coordination. These broader approaches to care coordination can facilitate the overall improvement of health care service delivery [3].

Health care professionals and older adults with chronic illnesses highlight the importance of participating in health-related decision-making and achieving self-management goals [13,14]. Receiving health-related information and managing one's health and care are important health-promoting activities that enable patient participation [14-16]. The Word Health Organization (WHO) defines participation as "being involved in a life situation" [17]. Thompson [16] describes 5 levels of patient involvement, from the lowest to the highest: (0) Noninvolvement: patients can lack medical knowledge, have low self-confidence, and receive care as passive patients; (1) Information seeking or information receptive: patients are provided information; (2) Information giving or dialogue: patients are listened to and heard, and patients and professionals share information; (3) Shared decision-making: patients make informed choices, receive guidance, and express their opinions in cooperation with professionals; and (4) Autonomous decision-making: patients can make choices independently, manage their health, and decide for themselves if they want to inform the professionals. Patients describe participation in managing their health as feeling respected, being heard, and being listened to by health care professionals who acknowledge their perspectives on their health situation [18].

A key purpose of participation is to provide relevant information to the patients according to their individual needs [18]. Despite a wide variation in the amount of health information needed by multimorbid older adults [19], many desire access to information about their health and some use eHealth to do so [4]. eHealth is defined as "the use of information and communication technologies (ICT) for health" [20]. A recent scoping review identified the following eHealth tools used in care coordination for older adults: patient portals, electronic health journals, telehealth monitoring solutions (sensor technology, virtual ward, and video consultations), and the use of the telephone [21]. These tools can ensure information sharing, enable communication between older adults and health care professionals, and support self-management of chronic illnesses [3,21-23].

eHealth can be an enabler for older adults to take an active role in their care [21,24,25]. Scholz Mellum et al [26] described how older adults with multimorbidity coordinate their care as they desire to be in control of their health. However, Elliot et al [27] reported that older adults living at home do not participate frequently in decisions regarding their health. Lack of time and difficulties in sharing and receiving information about patients were reported by the study participants (primary care nurses, social workers, and care coordinators) [27]. A review by Peart et al [28] showed that health care professionals assisted the self-management of home-dwelling older adults by actively involving them in care planning. The patients appreciated that their needs were respected by health care professionals [28].

Knowledge, confidence, and support from health care professionals and family members are factors that can enable the participation of home-living older adults in coordinating their care and their use of eHealth [21]. However, when supporting older adults' needs, health care professionals have found it challenging to incorporate older adults' health preferences [27,29]. Care coordination programs or interventions alone are insufficient to ensure the reduced use of health care [9]. The lack of incorporation of older adults' health preferences and the insufficiency of care coordination programs call for other approaches to ensure successful participation and eHealth use in care coordination. Therefore, this study aimed to explore multimorbid older adults' experiences with participation and eHealth in care coordination with general practitioners (GPs) and district nurses (DNs) [27]. The following research question guided our study: How do older adults experience participation in care coordination with GPs and DNs, and how does eHealth support their participation? This knowledge can allow primary health care services to develop strategies for delivering services that engage older adults, promote the self-management of chronic illnesses, and ensure the successful use of eHealth in care coordination [10,11,30].

Methods

Study Design

This was an explorative qualitative study conducted on health care services in a Norwegian municipality. Norwegian health care services are based on principles, such as universal coverage and equal access to health care services for all. The health care services are organized at a specialist and primary care level [31]. Norway, along with other Scandinavian countries, ranks

the highest in digitalization. For example, all Norwegian citizens must use electronic identification to access banking services [32]. In 2017, they were introduced to a personalized patient portal with the following features: an overview of medication prescriptions, appointments at the hospital or with GPs, access to hospital medical record systems, and the possibility of sending electronic messages to their GPs [33]. Primary care services are responsible for prevention, treatment, and early diagnostics. In addition, primary care services have financial responsibilities for citizens when hospitals consider them ready to be discharged to their municipalities [34,35]. Norwegian municipalities deliver home care services to 3.6% (203,000) of the residents in Norway, and the number of older adults receiving services is increasing [36]. Norwegian home care services can assist and educate community-dwelling patients in performing practical chores, such as cleaning the house, taking out the garbage, and cleaning clothes. Other services provided by home care services are health-related services in the home, such as district nursing services or providing day services for older adults to socialize, share meals, and be active [36]. All residents in Norway have the right to a GP. GPs are employed at different GP offices within the municipality, which are open during the day from Monday to Friday [37]. In addition, municipalities are responsible for emergency services, such as local emergency rooms that are open 24 hours a day, 7 days a week. Citizens in a municipality who need acute treatment are either referred to the local intermunicipal acute inpatient care (AIC) unit or the nearby regional hospital [37].

Study Setting and Participants

The study was conducted in a Norwegian municipality with 80,000 residents, where older adults live both in rural and urban areas. The district nursing services are organized into 5 different districts. The municipality has about 70 GPs who are co-located in different GP offices. The GP offices have nurses and administrative health personnel responsible for the telephone and for conducting clinical tests (eg, blood pressure examinations, blood tests, and spirometry).

We used convenience sampling to recruit older adult participants, and they were recruited either by their GPs or by a nurse working at the AIC unit [38,39]. The research team (HMHF and HB) reached out to each of the GP offices in the municipality and invited the GPs located there to participate in the study. We used the following inclusion criteria when recruiting the older adults: age >65 years, having two or more medical conditions, being on four or more medications, living at home, receiving district nursing, and having a hospital admission within the last 12 months before inclusion in the study. The GP or nurse at the AIC unit assessed if the included participants met the inclusion criteria. For older adults to be included in the study, all inclusion criteria had to be met. A total of 24 older adults were invited to participate, and 20 older adults were included. The reasons for declining to participate were not wanting the interview to be audio recorded and not finding the research relevant.

Data Collection

We used a semistructured interview guide developed by the research team (HMHF, HB, AM, and MS). The team consisted of researchers with competencies in social science, leadership, eHealth, general medicine, or nursing. The interviewed adults were asked about their age, gender, and medical conditions; the reason for hospital admission; what services they received from the municipality; and who followed their postadmission care. The interview guide focused on the following aspects related to experiences with care coordination: methods of communication with their GPs and DNs, use of eHealth, participation in decisions regarding their health, and efforts by health care professionals to involve them in coordination. We ensured that the older adults were given questions that were open-ended and that they provided examples of what they did in a situation (eg, what they did to manage medications or how they ensured that they understood the information given by health care professionals about their health or services). The interviews lasted between 30 and 90 minutes, and were carried out between October 2019 and February 2020. The first and third authors (HMHF and HB) conducted the interviews and had weekly meetings during the data collection period. The older adults were interviewed in their homes or at the AIC unit.

Ethical Considerations

This study is part of the research project “Leadership and Technology for Integrated Health Care Services” reviewed and registered with the Norwegian Agency for Shared Services in Education and Research (Sikt) (reference number: 228630). Sikt ensures data protection and legal access to handle research data. The research project was exempt from a formal review by the Regional Committee for Medical and Health Research Ethics in Norway (reference number: 2019/1138) because the project did not intend to generate further knowledge about health and disease. The research team (HMHF and HB) contacted the older adults after they provided oral consent to their GPs or the leader at the AIC unit to participate in the study. All participants were provided with written and oral information about the study. They were informed about the aims, methods, potential risks, and benefits of the research project according to the Helsinki Declaration [40]. Participants were informed that participation was voluntary and that they had the right to withdraw from the study at any time without giving any reason. The data were collected and stored according to data protection regulations.

Data Analysis

The interviews were audio recorded and transcribed verbatim. All personal and identifying information was removed to ensure the anonymity and confidentiality of the interview participants. NVivo 12 [41] was used to analyze the text. The analysis was guided by the 4-stage systematic text condensation approach described by Malterud [42] to present the data material as condensed text in categories with subcategories.

Stage 1

HMHF, HB, AM, and MS read all the data material and gained a general overview of the data associated with older adults’ experiences with participation and eHealth in care coordination with GPs and DNs. This resulted in the identification of the following preliminary themes: communication with health care professionals, experiences with the use of eHealth, information flow, and medication management. All the authors met in an

analysis meeting to discuss the data material and preliminary themes.

Stage 2

In the second stage of the analysis, units of meaning from the transcribed material were identified, and code groups based on the preliminary themes were established. HMHF coded the data and had several analysis meetings with MS and AMLH, who provided input on the codes and progression of the data analysis.

Stage 3

The code groups were grouped into 7 subgroups and reviewed. HMHF made a condensate illustrating each subgroup. The condensate was a summary of the original transcribed data material.

Stage 4

The subgroups were divided into 2 categories. We synthesized the condensate to text describing the categories and subgroups, and illustrated the text with quotes from the participants. Table 1 provides examples of the analysis process, with extracts from the data material.

Table 1. Examples of the systematic text condensation analysis process.

Preliminary theme	Meaning unit (quote)	Subgroup	Category
Medication management	The DNs had control of my medication before. But there were so many errors in the medication from a dispenser that I stopped the service. Firstly, some medications are missed. Then there was some medication I did not recognize. Then, I said, let me do it myself, administrate the medication. I take the medication in the evening and morning. [75-year-old woman]	• Managing own medication Excerpt from condensation of the subgroup: I am in control of my medications; I trust myself more than others. Even though I receive medication on a roll, I manage some medication myself. I adjust the dosage on my own. I have done that for many years, so I know what I am doing	Older adults in charge of and using eHealth in care coordination
Experiences with the use of eHealth	The GP has all information on the computer and can monitor it. They can bring out my whole history from the hospital. I feel confident that they know why I am there and will control my blood values and so on. [84-year-old man]	• Using eHealth to be informed or to seek health information Excerpt from condensation of the subgroup: The GPs and DNs communicate electronically, but they tell me if there is some information that's about me. I know that the hospital gives the GP electronic reports. This information is important to me and my GP. It gives me confidence that the GPs can access my electronic journal, especially if I am going to another doctor who is not my GP.	Older adults in charge of and using eHealth in care coordination

In the final stage of our analytic work we used a deductive approach. Thompson's 5 levels of patient involvement [16] were used to systematize the results to illustrate participation in care cordination among multimorbid older adults.

Results

Description of the Participants and eHealth Use

The mean age of the study participants was 82 years (range, 71-98 years). Of the 20 participants, 13 (65%) were female and 7 (35%) were male. As described in Table 2, the participants had various health problems, including chronic obstructive pulmonary disease, heart failure, chronic pain, cancer, physical disability, depression, and anxiety. A majority of the older adults in the study used the telephone to contact their GPs or home care nurses. Some had safety alarms, sent electronic messages to their GPs, or used electronic patient portals to share and access information about their health, such as appointments and prescriptions for their medications. A few interviewees used ICT, played games on their iPads or tablets, or paid their bills using their computers. Some had help from their caregivers to send text messages or call GPs or DNs. One participant was blind and was not able to access the electronic patient portal.

Table 2. Overview of participants' gender, age, self-reported health problems, and eHealth use.

Participant gender and age	Health problems	eHealth use
Woman, 82 years	COPD[a], heart failure, and chronic pain	• Uses the telephone to call GPs[b] or DNs[c] • Has a safety alarm
Woman, 98 years	Cancer and reduced mobility	• Uses the telephone to call GPs, DNs, or other health care providers • Has a safety alarm
Man, 71 years	Physical disability	• Uses the telephone and a tablet • Uses an electronic patient portal to control prescriptions and appointments, and to access the medical journal • Uses the telephone to call GPs • Receives text messages about appointments at the GP office or hospital
Man, 88 years	Chronic pain	• Uses the telephone to call GPs or DNs • Receives information from GPs via text messages • Has a safety alarm
Man, 84 years	Impaired eyesight	• Uses the telephone to call GPs or DNs • Uses a tablet to log into the electronic health portal • Makes appointments with GPs electronically
Woman, 81 years	COPD	• Uses the telephone to call GPs or DNs • Has a safety alarm
Man, 84 years	Blindness, diabetes, and physical disability	• Uses the iPhone and Siri functions to write text messages and call GPs • Has a safety alarm
Woman, 85 years	COPD, heart condition, diabetes, and femur fracture	• Uses the telephone to call GPs or DNs • Has a safety alarm
Woman, 85 years	Hypertension, metabolic syndrome, cancer wound, and femoral neck fracture	• Uses the telephone to call GPs or DNs • Has a safety alarm
Woman, 71 years	COPD, heart failure, depression, and chronic pain	• Uses the telephone, tablet, and computer • Has a safety alarm • Uses the computer to write online consultations with GPs • Uses Google to find information about treatments
Man, 80 years	No report of illness or health problems[d]	• Uses the telephone to call GPs or DNs
Man, 88 years	No report of illness or health problems[d]	• Uses the telephone to call the GP or DNs • Has a safety alarm
Woman, 75 years	Digestive issues and insulin use due to unstable blood sugar	• Uses the telephone to call GPs • Has a safety alarm
Woman, 87 years	No report of illness or health problems[d]	• Uses the telephone to call GPs • Next of kin helps with calling DNs • Has a safety alarm
Man, 80 years	No report of illness or health problems[d]	• Uses the telephone to call GPs • GPs can send text messages, including information on appointments • Has a safety alarm
Woman, 84 years	Anxiety and issues with pneumonia	• Uses the telephone to call GPs or DNs
Woman, 90 years	Chronic pain and bone fragility	• Uses the telephone to call GPs • Next of kin helps send text messages to GPs

Participant gender and age	Health problems	eHealth use
Woman, 72 years	COPD, paralysis due to cerebral stroke, and cerebral vision impairment	• Uses Google to get health information • Uses the telephone to call GPs or DNs, but often receives assistance from the next of kin • Has a safety alarm
Woman, 88 years	Hypertension, anxiety, and urinary tract issues	• Uses the telephone to call GPs or DNs • Next of kin helps send text messages to GPs • Has a safety alarm
Woman, 82 years	Chronic pain and cerebral stroke	• Uses the telephone to call GPs • Sends text messages to GPs

[a]COPD: chronic obstructive pulmonary disease.

[b]GP: general practitioner.

[c]DN: district nurse.

[d]Did not self-report health problems.

Older Adults in Charge of and Using eHealth in Care Coordination

The ability to communicate with GPs and DNs was central for the older adults to be in charge of coordinating their care. Managing their medications and using eHealth to be informed or to seek health information were aspects of engagement in care coordination.

Communicating With GPs and DNs

Communicating with GPs or DNs was essential for the older adults to be in charge of care coordination. To experience useful collaboration, they had to be able to talk to GPs and DNs, express their thoughts, and ask questions. Several interviewees talked about how they experienced collaboration with GPs as useful and how they could talk about their thoughts regarding their health. Furthermore, GPs educated them on health issues or treatments. Several participants had GPs who asked them about their opinions related to the treatment or follow-up of their health issues. A few of the older adults said that their GPs inquired about any other services and referred them to the health and welfare office or the hospital, if necessary. Many interviewees stated that they wanted to participate in and oversee their health. One older adult described that he was in control of his life and stated what the GP should do to help him:

> *I have taken the lead in my own life. I am in charge, not the GP or someone else or you. I am in charge and that's it! (...) That is how I want it to be to the bitter end. Because it is not the GP that should take the lead in the patient's life. It is the patients that live it themselves. (…) You [the GPs] should just provide the patient with tools.* [88-year-old man]

Many participants had monthly appointments with their GPs, which they booked in advance. This gave them an arena to communicate with their GPs and ask questions about issues concerning their medical condition or about the information they did not understand. The appointments with GPs were also important for giving assurance of follow-up on various tests. Some appreciated the opportunity to talk to their GPs as they had many health issues.

> *Sometimes the GP uses some expressions, and then I ask "What do you mean by that?" The GP could have said "today your blood values are fine", and your blood pressure is so and so. Instead of me having to ask and ask.* [81-year-old woman]

Several interviewees had daily visits by DNs, while others had weekly visits. Some personnel were nurse assistants, while others were registered nurses, responsible for tasks such as administering medication and injections. DNs assisted some of the older adults to get in contact with GPs, and also shared information with GPs about their medical conditions, medications, or health care appointments. Patients also used DNs for medical advice when they felt ill. DNs guided the older adults on what to do and performed measurements, such as blood pressure or blood sugar assessment, as illustrated by a participant:

> *I have asked the DNs if they can come and test my blood sugar since the blood sugar sometimes has too high levels.* [85-year-old woman]

DNs were described as being nice. They were people to talk to about daily life and were always available for support. A few participants perceived DNs as having experience with caring for patients with reduced mobility and having knowledge on how to help them. Some interviewees perceived that DNs were updated on their situation and could inform them about new medications:

> *Yes, the DNs will always stop by the day I arrive home [after being transferred home from the hospital]. If there are changes in my medication, the DNs are updated and inform me about the new medication.* [82-year-old woman]

Managing Own Medications

Managing medications was an important aspect of older adults' involvement in care coordination. Many of the older adults participated in dispensing their medications from multidose dispensers, which informed them of the date and time to take the medications. Some also had other medications on the side that they managed themselves, sometimes in dialogue with their

GPs and sometimes without involving their GPs. A few took paracetamol (acetaminophen) when they had pain and wanted to manage the pain medication themselves. One of the female participants explained that she discussed her medications with the medical doctors at the AIC unit and that they had decided that it was safe for her to be in charge of the pain medications herself.

A few interviewees explained that they trusted themselves more than health care professionals when it came to taking the correct medication and dosage at the right time. In addition, they received the information they needed from the pharmacy about side effects, and when and how to take the prescribed medication. According to these interviewees, this information was not given by DNs or GPs. Some participants experienced changes in their medication after being discharged from the hospital, or when they received a multidose medication dispenser. They remembered what medication they were supposed to take, and in that way, they made sure that the medication was as prescribed. One woman explained that she found errors in the multidose medication dispenser and that she stopped the service and took control of the medication management herself:

> *The DNs had control of my medication before. But there were so many errors in the medication from a dispenser that I stopped the service. Firstly, some medications are missed. Then there was some medication I did not recognize. Then I said, let me do it myself, administrate the medication. I take the medication in the evening and morning.* [75-year-old woman]

Use of eHealth to be Informed or to Seek Health Information

Access to eHealth ensured that older adults could participate in care coordination by obtaining or seeking information over the telephone, patient portals, or the internet. Although all the interview participants had access to their patient portals, they rarely took advantage of this opportunity. Most of the older adults included in the study were able to use the telephone to contact their GPs, DNs, or the hospital. A few interviewees said they did not like to use the telephone to call GPs or DNs, but they were able to use the telephone to contact their family members or friends. Some explained that if there were test results or some health-related information they did not understand or needed more information about, they could call the GP office and ask questions. Even though they did not reach the GPs when they called, the nurses knew who they were and ensured that they got an answer from the GPs. Some interviewees said they did not feel the need to read their medical records or log into their patient portals, as they preferred to talk to their GPs or DNs face-to-face. A few did not know they could access the electronic journal from the hospital through a patient portal.

> *Do we have that (online access to their electronic journal)? I have not tried to get hold of that information (the electronic journal), and I have not had the need. I have been informed enough as it is kind of. I can ask if there is something I wonder about.* [82-year-old woman]

A few older adults used the patient portal to be updated on appointments or medication prescriptions, or to send electronic messages to their GPs. They pointed out that they felt secure knowing that their GPs could always access the information about them, as information from the hospital is transferred electronically to their GPs.

> *The GP has all information on the computer and can monitor it. They can bring out my whole history from the hospital. I feel confident that they know why I am there and will control my blood values and so on.* [84-year-old man]

Some interviewees explained that their GPs called them to inform them about test results, even though that happened rarely. Only a few participants used the internet to gain information about their health conditions and find possible treatments that they suggested to their GPs.

> *Yes, she (the GP) listens to me. The last time I was there with heart failure, I Googled and then I read something that perhaps it can be surgically treated. Something like a pacemaker or a heart starter. Yes, for heart failure it said.* [71-year-old woman]

One interviewee pointed out that older people need to be taught how to use ICT, making sure that older adults can be updated on available eHealth services.

> *People are getting older and older, and policymakers cannot expect people to use everything on the computer and digitally. They must teach the older group too… so that people are updated.* [90-year-old woman]

Older Adults With a Loss of Control in Care Coordination

Being dependent on guidance from GPs and DNs, and lacking the capacity to be involved in information sharing were aspects resulting in a loss of control in care coordination.

Dependence on Guidance and Help From GPs and DNs in Managing Health

Some of the interviewees experienced challenges that limited their participation in care coordination. Some participants lacked health literacy and needed help managing their medications. Several of the older adults talked about respecting and trusting the GP's knowledge. They perceived that the GP had the best knowledge about their health problems and how to treat them properly. Some explained that they needed help from GPs and DNs to manage their medical conditions. Receiving health care services was necessary to manage their health. Therefore, they followed the instructions and plans of the GPs on how to self-manage their medical conditions. Seemingly, some older adults lacked sufficient health literacy to know what is best for them and had to trust GPs:

> *I don't know what is best for me. It is the GP that suggests different solutions. I follow the GPs advice*

> *without thinking more about it. I trust the GPs assessment.* [84-year-old man]

Some of the interviewees needed more help managing their medications. DNs helped some patients to remember to take their medications and signed on a sheet of paper when they had taken the medication. Other participants had reduced physical function, and some explained that GPs or DNs had to assist them in administering eye drops or taking medications at the right time. One female participant also said that she wanted to put the medication in the dispenser herself, but she had difficulties with her fine motor skills and needed assistance from DNs.

> *In the beginning, I put the medication in the medication dispenser myself. I do not do that anymore. It is a long time ago. I miss it because I like to be in control.* [90-year-old woman]

Lack of Capacity and System Support to be Involved in Care Coordination

Some older adults could not remember relevant health information and experienced difficulty becoming involved in care coordination. A lack of system support also hampered participation. Many of the interviewees perceived that they did not remember all information regarding their health after being discharged home from the hospital or AIC ward, or after meeting their GPs. They were too sick to be able to remember all information. A few interviewees experienced the information sharing with DNs as challenging. Having several DNs or nurse assistants coming to their homes made it hard to get to know those working in the district nursing service. Therefore, some participants did not share important information, such as information on recent hospital admissions or hospital discharge letters, because of a lack of continuity with DNs.

> *I don't talk with the DNs. I don't even mention that I have been hospitalized. I think they know, but I can't relate to them as there always is a new DN stopping by. If there had been one permanent nurse, I could at least talk with them about stuff, but no, it is not like that. They just give me my medications and say goodbye.* [81-year-old woman]

A few participants described a lack of collaboration with their GPs due to the infrequency of visits to the GP office. They said that GPs lacked information about their current health status, as illustrated by a female participant:

> *I think there has been very little collaboration with the GP. He called me, and then I had an appointment with him right after I was discharged home from the AIC unit. So, I have visited the GPs office, but it seems he does not have a full understanding of what has happened to me. He kind of ask me things I thought he should know.* [72-year-old woman]

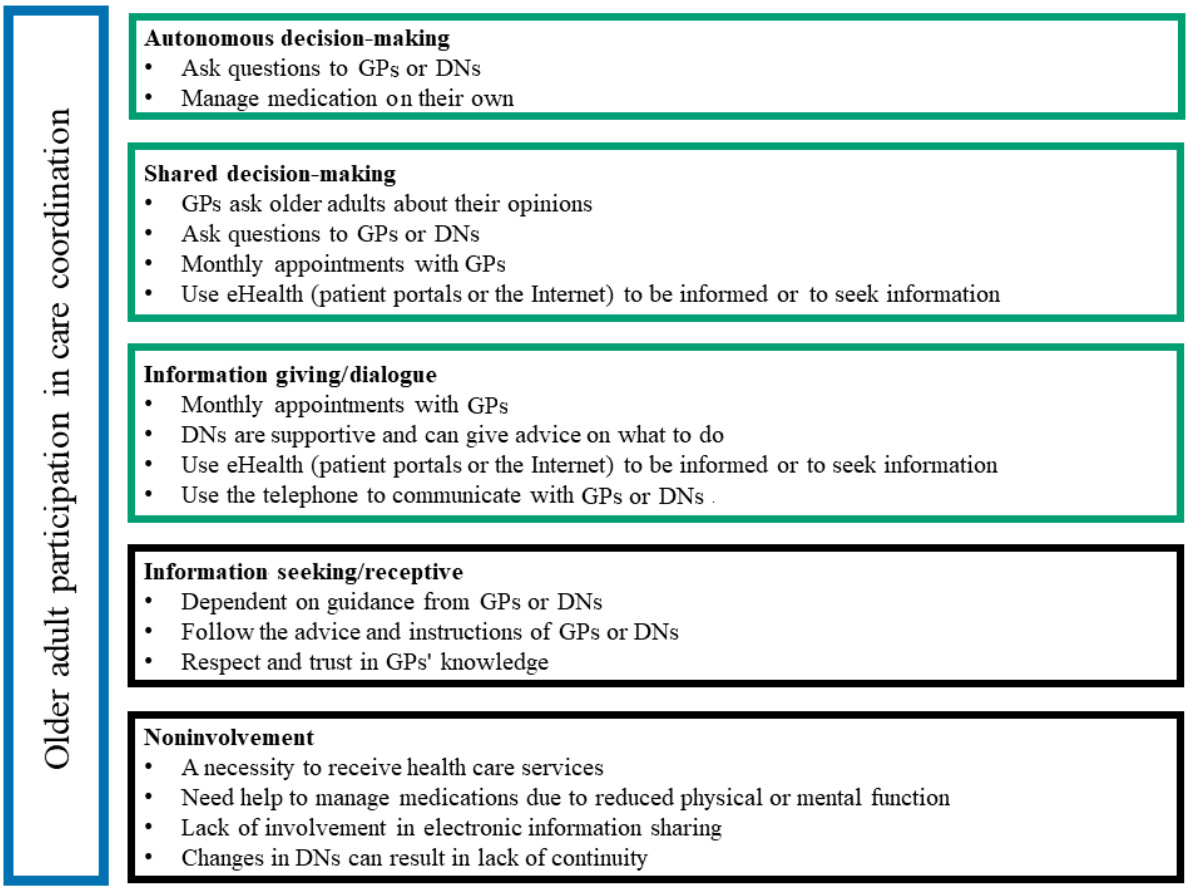
Some older adults experienced not being involved in electronic information sharing between DNs and GPs. They did not know what was written in the electronic messages exchanged between DNs and GPs. One participant said:

> *I don´t like it. They can ask me. They can come to me and ask me directly and talk to me before they write something (in the journal). But they don´t do that.* [90-year-old woman]

Participation in Care Coordination Among Multimorbid Older Adults

There were 2 categories of older adults: (1) older adults in charge of and using eHealth to coordinate their care, and (2) older adults with a loss of control to coordinate their care [16]. By using the 5 levels of involvement by Thompson [16], we systematized the results impacting participation in care coordination for older adults (Figure 1). In Figure 1, the first category has been visualized in green text boxes as higher levels of patient involvement [16]. Older adults who manage medication and ask questions to their GPs or DNs can achieve autonomous decision-making. Shared decision-making is facilitated by GPs who ask patients for their opinions, older adults who use eHealth to be informed or to seek new information, and monthly appointments with GPs. Monthly visits with GPs provide an arena for dialogue, in line with level 2 of patient involvement [16], which includes the use of the telephone to communicate with GPs and DNs, and a sense that DNs are supportive. The second category has been visualized in black text boxes as lower levels of patient involvement [16]. Some older adults have a low level of participation as they follow guidance from their GPs, have high respect for the GPs' knowledge, and need help managing their medications. Other aspects impacting a low level of participation include a lack of mental or physical function to manage one's medications, necessitating assistance from DNs and health care services. Changes in DNs can result in a lack of continuity since it is difficult for older adults to get to know them. This can cause a low level of patient participation. In addition, a lack of involvement in electronic information sharing can hamper older adults' participation in coordinating their care.

Figure 1. An overview of participation in care coordination among multimorbid older adults in a primary care setting. Some aspects were common for autonomous and shared decision-making, and for shared decision-making and dialogue.



Discussion

Principal Findings

The study aimed to explore multimorbid older adults' experiences with participation in care coordination, and how eHealth may support participation. The results suggest that older adults have various experiences with participation, communication, and collaboration with their GPs and DNs, and in the use of eHealth. It is important for older adults that GPs ask them about their own opinions and that they have DNs who are supportive of managing their health. Furthermore, managing medication and using eHealth to seek information can ensure high levels of participation in care coordination. All participants called friends, family members, GPs, or DNs, indicating that the telephone is a simple and efficient means of ensuring patient involvement. However, issues related to involving older adults in care coordination remain, as some older adults perceived that their GPs or DNs did not collaborate much and did not always know what had happened during the last hospital admission. Some participants indicated that they respected their GPs' knowledge and followed their GPs' or DNs' guidance and instructions. Lacking the capacity and system support to be involved made older adults lose control in care coordination. Further, reduced physical capacity to manage medication without help or difficulty remembering all information made it challenging for older adults to participate. The results pinpoint different levels of participation of multimorbid older adults in care coordination and eHealth use. This can provide health care professionals and patients with crucial knowledge of what to be aware of or what options there are when multimorbid older adults become involved in care coordination and use eHealth successfully.

Levels of Older Adults' Participation in Care Coordination

Our results showed that some older adults participated in care coordination, and a strong relationship with GPs seems to facilitate a high level of involvement. A strong relationship of patients with GPs and DNs can be characterized as older adults asking questions to their GPs or DNs, having monthly appointments with their GPs, DNs being supportive, and GPs asking the older adults for their own opinions. A strong relationship of older adults with their GPs and DNs is an enabler for participation in the care coordination process [3,18,28,43]. Our results also showed that having monthly appointments with GPs provides an arena to talk and ask questions. Scholz Mellum et al [26] found that older adults with multimorbidity often had regular appointments with their GPs to manage their health. Having regular appointments with GPs can facilitate a strong relationship between GPs and patients, and increase the probability of participation in their care and health [26]. Steele Gray et al [43] identified that health care professionals need to recognize community-dwelling patients with complex chronic illnesses as whole persons by listening to them and having a strong relationship with them. A recent systematic review found that continuity of care with GPs or hospital doctors was

associated with lower mortality rates [44], highlighting the necessity of having a strong relationship with GPs. Parisi et al [45] reported that high turnover among GPs reduces patients' experiences of continuity of care and is linked to poorer services and patient health outcomes. Findings from our study indicate that DNs can have a good relationship with older adults, being perceived as supportive in coordinating care. Nilsen et al [46] reported that older adults who had recently been discharged from the hospital to home characterized some DNs as marvelous and kind, as well as supportive in coordinating their care. DNs can have a central role for older adults, which is especially important if GPs are not able to follow discharged patients' health and care.

Our results show that some older adults are able to take their medications themselves, taking control of the medication management and thus making an autonomous decision about their care [16]. Managing medication is important in care coordination, especially in reviewing medication regimens or assessing all medications or supplements a patient takes [3]. We found that some older adults need assistance from DNs because of reduced physical capacity to manage their medications. It is unclear if this is something the patients want themselves. Older adults wish to be in control and to be as independent as possible, despite reduced functional ability [47]. Other research has found that older adults find it important to be informed and feel in control of their medications [48,49], and being informed can contribute to feeling in control and ensuring that information giving, dialogue, and shared or autonomous decision-making occurs [16]. In a qualitative study of 19 older adults with multimorbidity, Löffler et al [50] found that they often were critical about their medications. In a mixed method study of older adults with polypharmacy (prescribed five or more medications), Clyne et al [51] found contrasting views on taking medication, where one group of older adults believed strongly in the medication and another group were concerned about adverse effects. Similarly, we found that there are mixed experiences when older adults manage their medications. For those who are not able to manage their medications themselves due to reduced physical or mental capacity, DNs play an important role in supporting them. Robinson et al [52] highlighted the pivotal role of DNs in following up on medication and helping older adults to adhere to the medication regimen. However, our results show that some older adults trust themselves more than DNs when managing their medications. Similar results were reported by Schiøtz et al [53], with study participants not feeling confident that DNs had the necessary information about their medications. Schiøtz et al [53] highlighted that having a physician coordinating their care made the included patients feel secure about the care and medication they received. Our results suggest that for some older adults, DNs are supportive and assist them in managing their medications.

Our findings show that older adults with multimorbidity have different experiences engaging in their health. Some are in charge, while others have lost control of care coordination. According to the 5-level ladder of involvement by Thompson [16], noninvolvement is the lowest level, in which patients do not wish to be involved in discussions regarding their health. Our results indicate that patient preference is not the only factor. Noninvolvement can also be explained by a lack of system support, and GPs and DNs not having sufficient information about older adults' health and therefore not supporting them. Other aspects impacting noninvolvement include reduced physical or mental capacity. However, according to Thompson's description of being information seeking or receptive, patients trust the information given by health care professionals without really being involved [16]. Similarly, our results show that some older adults seem to respect GPs' knowledge and advice so much that it hampers their participation. This is also reported in other studies involving older adults [26,49,54,55], especially those of advanced age [49]. Some older adults accept that decisions are made for them [54] and follow recommendations from physicians or nurses [26,55] without asking questions. Osborn et al [56] also found that health care professionals often neglect to include older adults with chronic illnesses in decision-making regarding their health. The study identified a lack of information sharing and talking with older adults about what they could do to promote their health [56]; this can act as a barrier to both care coordination and participation. Oksavik et al [57] also found various levels of participation when older adults with multimorbidity engaged in care planning meetings with health professionals. Thompson pointed out that the different levels of involvement are dynamic and that patients can move between levels [16]. For example, being too sick after a hospital admission makes it challenging for older adults to remember all information regarding their health and participate in coordinating their care. In a situation of acute illness or worsening of chronic illness, older adults move to a lower level of participation [16], impacting their capacity to manage care coordination. Health care professionals aware of this can provide extra education or advice to older adults in acute situations. Doing so can ensure that patients can easily self-manage their illnesses and navigate health care services, thus reducing health care use [9,10].

Role of eHealth in Older Adults' Participation and Care Coordination

Our results showed that some older adults use eHealth tools to communicate, seek information about their health by using a patient portal, find information on the internet, or send electronic messages. The study participants were able to use the telephone to communicate and be informed about their chronic conditions with GPs, DNs, or nurses at the GP office. Fjellså et al [21] reported that the telephone is one central eHealth tool that facilitates care coordination for older adults living at home. Oh and Lim [58] found that older adults who experienced negative communication with health care providers (ie, not receiving sufficient information on a health issue) started to search for information on the internet. However, those study participants did not have multimorbidity and were experienced with using the internet [58]. Some older adults in our study were able to use the internet to search for information, access their electronic patient portal, or send electronic messages to their GPs; this was an enabler for participation. These older adults had a degree of eHealth literacy, defined as "the ability to seek, find, understand, and appraise health information from electronic sources and apply the knowledge gained to addressing or solving

a health problem" [59]. Previous research [21,60] has indicated that adults who are very old (ie, aged 80 years or over) have limited use of eHealth. Some of our study participants were over 80 years old and used eHealth. This may indicate that eHealth literacy can be found in individuals of advanced age. Other older adults did not use eHealth but were able to use their computers or tablets to pay bills digitally or play games. Older adults can have computer literacy without having eHealth literacy [59]. As 1 participant reported, older adults need to be educated to enable the use of eHealth. Research has shown that sufficient technical support and education are essential to increase eHealth use and health literacy among older adults [21,61-63]. Increasing eHealth literacy can ensure that older adults can use relevant information to self-manage their chronic illnesses as part of care coordination, which is an important enabler for both participation [17] and health promotion [15].

Strengths and Limitations of the Study

Our study included 20 participants recruited from different GP practices in a Norwegian municipality, which ensured various patient characteristics and experiences in several aspects (participation, care coordination, and eHealth use) [64]. The recruitment of older adults may have been impacted by GPs' or nurses' busy work schedules and their subsequent capacity to recruit. They may also have perceived that the patients lacked the ability or interest to participate [65]. There was variation in what older adults said in the interviews; some did not provide long descriptions, while others gave well-articulated descriptions of their experiences. By including 20 older adults in the study, we were able to identify the variation among the participants, and this ensured that we met sufficient information power for the aims of our study [64]. The use of a convenience sampling strategy may also have led to missing experiences and opinions from older adults with extensive experience with eHealth and participation in care coordination [39]. However, we included older adults with multiple chronic conditions who were living at home and who were very sick and had excessive health care use. They can be "hard-to-reach" research participants; thus, this study addresses the needs and knowledge of a vulnerable group of older adults [66]. This knowledge will benefit both the general population and older adults with and without extensive experience with eHealth and participation in care coordination.

In this study, we included only experiences regarding health care services, not services from community social care and mental health. However, we included questions to map the sources of services, and no participant mentioned other services than health care, even though some mentioned having issues with depression or anxiety. Future research should highlight both mental health and social services for older adults living with multimorbidity.

Relevance to Clinical Practice

Based on our results and discussion, participation in care coordination among older multimorbid adults, which supports the self-management of chronic conditions, can be improved by ensuring communication, facilitating management of one's own medications, promoting a strong relationship with health care personnel, encouraging use of eHealth for information, and educating on eHealth tools. Health care professionals in charge of older adults' care should attempt to map the individual's willingness to participate in care coordination and to allow them to speak their minds. This is perhaps particularly applicable to GPs, as this study shows that GPs appear to have an important role in care coordination for older adults with multimorbidity. Many of the participants in this study wanted to manage their medication themselves, and supporting older adults to do this is important, despite any reduced physical ability. eHealth has shown promise as a tool to improve older adults' participation in decision-making concerning treatment and care coordination [16]. However, older adults must be given the opportunity and education to use eHealth to participate in coordinating their care. The use of the telephone with GPs or DNs to share information, ask questions, and support self-management of chronic illnesses is important for older adults. Future research should increase knowledge of how health care providers support participation and use of eHealth in care coordination for older adults with multimorbidity. It is especially relevant for future research to achieve a better understanding of electronic information sharing between GPs and DNs, as this can trigger various levels of patient participation. Thus, promoting and sharing information about available eHealth tools, and electronic information sharing with patients and personnel are important.

Conclusion

In this study, we explored older multimorbid adults' experiences with participation in care coordination and their use of eHealth to support such participation. High levels of participation include aspects such as communication, asking questions to GPs or DNs, managing one's medication, being asked questions by GPs, and using eHealth. Reduced physical capacity or reduced capacity to remember information and lack of system support can make it difficult to participate in all aspects of care coordination. The study findings are connected to Thompson's 5 levels of involvement [16], providing avenues for future research, and giving practitioners and policymakers a better understanding of how to increase older adults' participation in care coordination. Future research should contribute to a better understanding of electronic information sharing among health care providers because older adults may experience a lack of involvement in information sharing, thus hampering participation. Moreover, the results indicate that there is a willingness among older adults to use eHealth to participate in decision-making and to self-manage the coordination of their care.

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Authors' Contributions

HMHF, HB, AM, and MS designed the research project and developed the interview guide. HMHF and HB recruited participants and conducted interviews. HMHF analyzed the data with input from AMLH, HB, AM, and MS. HMHF took the lead in drafting, writing, and revising the manuscript. AMLH and MS participated in writing, commenting on, refining, and revising the manuscript. All the authors read, commented on, and approved the final manuscript.

Conflicts of Interest

None declared.

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Abbreviations

AIC: acute inpatient care
DN: district nurse
GP: general practitioner
ICT: information and communication technologies

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