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Viewpoint

Patient and Family Participation in Clinical Guidelines Development: The Cystic Fibrosis Foundation Experience

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Abstract

Patient and family participation in guideline development is neither standardized nor uniformly accepted in the guideline development community, despite the 2011 Institute of Medicine’s Guidelines We Can Trust and the Guideline International Network’s GIN-Public Toolkit recommendations. The Cystic Fibrosis Foundation has included patients and/or family members directly in guideline development since 2004. Over time, various strategies for increasing patient and family member participation have been implemented. Surveys of recent patient/family and clinical guidelines committee members have shown that inclusion of individuals with cystic fibrosis and their family members on guidelines committees has provided insight otherwise invisible to clinicians.

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KEYWORDS

cystic fibrosis; guidelines; patient participation; patient engagement

Introduction

Cystic fibrosis (CF) is a rare, genetic, life-shortening disease that impacts approximately 35,000 people in the United States [1]. The small population size has resulted in a paucity of evidence addressing many aspects of CF care, which impacts the development of clinical practice guidelines. Cystic Fibrosis Foundation (CFF)-sponsored guidelines bolster limited evidence with clinical and patient expertise. Since its first guideline was published in the peer-reviewed literature in 1992 [2], the CFF has continued to sponsor guidelines to standardize care and improve outcomes for individuals with CF. Guidelines are developed by committees of experts, including members of the CF multidisciplinary care teams and others who treat people with CF. For over a decade, CFF guidelines committees have also included individuals with CF and/or their family members. As experts in life with the disease, they provide essential information about their perspectives and experiences and can provide insights otherwise invisible to the clinical community.

To ensure that the outcomes of the CFF’s work fit within its chronic care model [3], is rooted in patient-centered needs, and recognizes the importance of community and patient engagement [4-6], the CFF has formalized the process of partnering with and including individuals with CF and their family members in guideline development [7,8]. The first individuals with CF and family members included on guidelines committees not only provided details on their lived experience with CF, but also brought their professional experiences in law, quality improvement, and medical writing to the committee. At this time, the CFF was undergoing a cultural shift towards an increased value in partnership between patients, families, and clinicians. As the culture at CFF continued to move towards partnership, those included on guidelines committees began to represent a broader segment of the CF community. A network of CF community members called Community Voice was developed in 2014 to assure that opportunities to participate are available to a broad spectrum of participants. This group, made up of individuals with CF and family members, is involved in shaping programs and initiatives that impact the broader CF community and has helped the CFF foster meaningful engagement and partnership with those it serves [9]. The development of Community Voice has enabled a more extensive range of patients and family members to participate in, or apply for, opportunities to partner with clinicians on guideline development.

Patient and Public Involvement in Guideline Development and Implementation

The Institute of Medicine's (IOM) standards for guideline development, published in 2011, call for the inclusion of patient and public involvement (PPI) [10]. The Guidelines International Network (GIN) toolkit, G-I-N PUBLIC, outlines different methodologies of PPI: Participation, Consultation, and Communication [11]. In 2011, a data synthesis found that of the 71 guideline manuscripts reviewed, 39% included PPI in a guidelines working group, 10% in the literature review, 34% in a consultative capacity, and 13% in a public poll or survey [12]. The best method for including PPI in guideline development and implementation has not been determined [13,14].

Below is a description of how the CFF involves the community in the development and implementation of guidelines and how clinicians and community members perceive the impact of their involvement using GIN's PPI methodology. The learnings from the CFF presented below could be adapted by other guideline developers to incorporate PPI into their process.

Participation

A recent study comparing parallel groups, one including patient representatives and the other not, concluded that PPI should be an essential part of trustworthy guideline development [15]. Others have argued that the participation of patients and families in all aspects of guideline development is not essential as long as their voice contributes to the determination of key questions addressed by the guidelines [16]. CFF guideline committees look to patient/family participants to provide insight into the priorities and perspectives of individuals with CF, to determine the topics addressed, and to weigh in on the recommendation statements. The inclusion of an individual with CF and/or a family member also offers insight into the real-world implementation of guideline recommendations from their lived experience with the disease. Including an individual with CF and/or a family member of someone with CF rather than a third-party patient advocate, as the community representative on the committee, highlights aspects of the lived experience of which clinicians and patient advocates may not otherwise be aware. The addition of patient preferences through the inclusion of PPI on the committee can inform the guideline development process [17,18], yield a more patient-centric and evidence-based guideline, and may increase care partnerships and the ability to sustain daily care [19].

While the IOM and GIN recommend PPI, a 2017 study found that just 8% of the 101 guideline developers reviewed require PPI on guideline development committees [20]. A 2008 study found 39% (12 of 31), and a 2012 study found 16.7% (19 of 114) of guideline developers included in the study used the participation strategy of PPI in the guideline development group [21,22]. Since 2004, individuals with CF and/or family members have directly participated in the development of all 28 CFF care guidelines by serving on specific guideline committees. Patient and family committee members work with clinicians to develop PICO (person, intervention, comparison, outcome) questions, are encouraged to participate in the literature review with guidance from other committee members, take part in drafting recommendation statements, and vote alongside other committee members on the final recommendation statements. They are encouraged to share their expertise from living with CF and experience with the guideline topic.

Consultation

Studies have shown that 33% to 45% of guidelines undergo an external review or public comment period that includes patients or general public commenters [20,21]. Starting with the Infection Prevention and Control Guidelines, published in 2014, the CFF has sought public comment on its draft guidelines, including feedback from individuals with CF and their families (Table 1). Before the initiation of Community Voice, draft guidelines were distributed to patients and family members through CFF multidisciplinary listservs, some of which included members of the patient and family community. After the expansion of Community Voice in 2017, the guidelines public comment period became more accessible to individuals with CF and their families. Public comment periods for new guidelines were consistently shared for international distribution in both the clinical and patient/family communities beginning in 2019. Two joint CFF and European Cystic Fibrosis Society guidelines, published in 2015 and 2016, had previously been shared internationally.

Table 1. Patient/Family Public Comment Feedback. Starting with the CFTR Modulator guidelines, published in 2018, the guidelines have been distributed through wider communication channels, including Community Voice. Since than the number of patient/family responses has increased.

Guideline (year published)	Individual with CF Responses	Family Members Responses	Total Individual with CF and Family Members with CF Responses to Public Comment
Infection Prevention and Control (2014)	5	9	14
Eradication of Initial Pseudomonas (2014)	2	1	3
Depression and Anxiety (2015)	14	17	31
Nontuberculous mycobacteria (2016)	2	6	8
Preschool Aged Care (2016)	0	16	16
Enteral Tube Feeding (2016)	4	7	11
Diagnosis (2017)	0	5	5
Colorectal Cancer Screening (2018)	2	3	5
CFTR Modulator (2018)	18	30	48
Lung Transplant Referral (2019)	23	11	34
Advanced CF Lung Disease (2020)	19	10	29
Models Palliative Care (TBD)	17	19	36
Post Lung Transplant (TBD)	18	5	23

Targeted surveys have also been used to obtain broader feedback from the individuals living with the disease. In 2017 and 2018, the CFF conducted surveys of individuals with CF, family members, and health care providers to inform the scope of upcoming guidelines [23-25]. These surveys provided insight that was otherwise unknown to the clinical committee members and informed the scope of the guidelines.

In 2017, the CFF piloted a focus group engagement strategy for the development of one guideline. This pilot included spouses and individuals with CF in a focus group lead by the psychiatrist and the adult with CF on the guideline committee. The focus group was able to provide insight from their experience to help to fill a gap in the CF-specific literature [26]. This supplemental group allowed the committee to hear from multiple individuals with CF and spouses of adults with CF, ensuring that broader perspectives and experiences informed guideline development. The input of the patient and family members on this focus group informed the guideline committee of previously unknown psychosocial barriers that contributed to suboptimal outcomes.

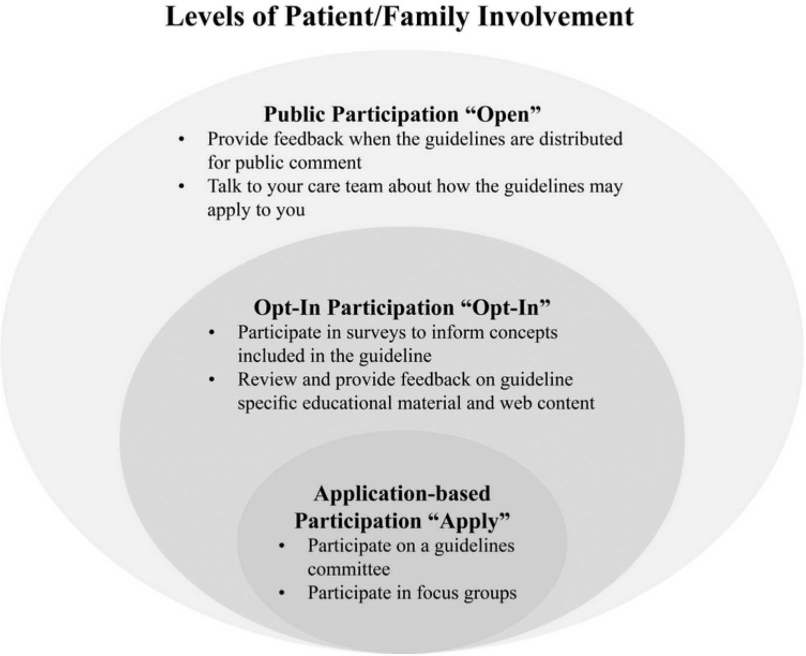
Communication

Since 2014, the CFF has developed public-facing material to support its guidelines. Previously, materials assisting clinicians in the implementation of these guidelines were developed and shared by the members of the guideline committee via listservs with other clinicians. Individuals with CF/family members on the committee are asked to provide feedback on the resources created to ensure a wider understanding of the guidelines, or other patients and family members are recruited through Community Voice. These resources are disseminated through emails to CF Care Center staff, CFF email listservs, Community Voice, and are posted to the CF Foundation website [27].

Overview

Guideline development via a partnership with clinicians, individuals with CF, and families enables productive interactions between care teams and patients. The concepts laid out by the GIN toolkit, participation, consultation, and communication, provide substantial opportunities for patients, families, and care providers to implement the chronic care model [28] and improve health outcomes. While CF care is provided within the framework of the chronic care model [3], the concepts presented here can be adapted to other patient groups. Before the CFF had access to the active patient and family population through Community Voice, many of the patients and families included in CFF guideline development were recruited by clinicians serving on the guideline committees. An overview of opportunities for patients and families to participate in CFF guideline development and implementation are outline is provided in Figure 1.

Figure 1. Opportunities for PPI in CFF Guidelines and Guideline Implementation: There are three levels at which individuals with CF and family members can participate in guideline development and implementation. 1. Anyone: these opportunities are open to all patients and families. 2. Opt In: These opportunities require patients and families to sign up for Community Voice to learn about these methods of influencing the guidelines and guideline implementation. 3. Apply: These opportunities require individuals who have opted-in to Community Voice to apply to participate in these projects.



Levels of Patient/Family Involvement

Public Participation "Open"

- Provide feedback when the guidelines are distributed for public comment
- Talk to your care team about how the guidelines may apply to you

Opt-In Participation "Opt-In"

- Participate in surveys to inform concepts included in the guideline
- Review and provide feedback on guideline specific educational material and web content

Application-based Participation "Apply"

- Participate on a guidelines committee
- Participate in focus groups

Challenges

Current CFF guideline committees are encouraged to include two individuals with CF on each committee, in addition to at least one family member, ensuring the inclusion of more than one individual's perspective and preventing the members with CF from feeling that they must represent the entire patient population. It enables the voice of an individual with CF to be present even if the other individual with CF becomes too sick to participate. However, including multiple individuals with CF has unique challenges. The CFF Infection Protection and Control guidelines, and CFF policy, recommend that only one person with CF attends any CFF-sponsored indoor event to decrease the potential for cross-infection [29]. The CFF uses virtual meeting platforms to enable the participation of more than one individual with CF.

The CFF's relationship with individuals with CF and their family members has been increasingly cultivated with the development of Community Voice. Individuals with CF and family members are now able to apply to participate in CFF projects like guideline committees. By 2019, it had over 1150 members, with participants from all 50 states. While there is broad representation in Community Voice, it does not reflect the entire CF population, as people must sign up to become a member. Members choose what types of projects they would like to hear about and participate in based on their interest and level of time commitment [30]. "Opt-in" membership, and recruitment options, can place limits on the variety of patient and family experiences and perspectives participating in guideline development and implementation.

Proactive partnerships between clinicians, patients, and families may lead to more actionable recommendations at the point of care. However, research is needed to fully understand the influence of PPI on the actionability of guidelines. Research on the effect of including patients and family members on the review of educational materials is needed. Understanding how PPI influences these factors is necessary to improve the guideline and related materials development processes.

Impact

While systemic reviews exist on the impact of patients in the setting of advisory councils [31], there is limited systematic evidence of the impact of the presence of an individual or family members on the formation of guidelines. This report has highlighted many areas where the input of individuals with CF or their family caregivers improve the focus and patient-centeredness of CFF guidelines. A critical factor in the guideline development process is the interchange between researchers and clinicians with individuals with CF and family members in real-time as PICO questions are created, the evidence is evaluated, and the outcomes are determined. The presence of an individual or family member living with the disease transforms the process from an academic exercise into a meaningful exploration of those questions and outcomes that have an impact on their daily life. These discussions also branch into areas that might not have been considered without the presence of these individuals.

Despite the limited published evidence of the impact of PPI on guideline development, the authors believe that including patients and families in guideline development has improved the guideline development process. In order to better assess the impact of this process of inclusion, in July 2019, the CFF surveyed clinical committee members who participated in a recent guideline committee that included at least one individual with CF or a family member of a person with CF. Fifty-seven individual responses were obtained from the 176 non-patient/family guidelines committee members (32%

response rate). Ninety-three percent of respondents agreed or strongly agreed with the statement that the presence of a person with CF or a family member of a person with CF improved the guidelines formation process. Sixty-three percent of respondents agreed or strongly agreed that the involvement of these individuals improved the PICO questions chosen, and 89% agreed or strongly agreed that the presence of these individuals improved the selection process for outcomes considered to be important. The survey also asked whether the presence of individuals with CF and their family members would constrain discussion. Only 9% of respondents agreed or strongly agreed that this was an issue.

An open-ended question in the survey asked the clinical guidelines committee members to describe what they found helpful about the inclusion of a person with CF and/or a family member of a person with CF on the committee. Of the 57 responses, 56 were uniformly supportive of the role. The responses were typified by one committee member who stated, “I learned so much from their presence. I valued their involvement very highly. I was able to ask them questions that I never really thought to ask patients before, and the experience was incredibly informative. Also, it helped shape my ideas of what questions we should be asking and how we should be tailoring care in consideration of how patients are directly impacted.” Another prevalent observation is summed up by another participant: “This was my first experience having a patient representative to help guide professional questions and decision-making. It was extremely valuable and provided a real-world representation of the needs in patients with CF who suffer with chronic medical issues. It was also enlightening to have the adult with CF indicate how her drug-induced hearing loss has affected her life and how she wasn’t provided much information or guidance about this risk during her treatments. I think having both a parent representative and an adult patient with CF was critical to keep the focus on ‘patient needs’ rather than ‘clinician wants’ during our discussions.” According to this participant, the patient representative on this particular guideline committee helped guide the PICO question development process by sharing a personal experience and how it impacted her CF care.

While some data can be found in the literature about the clinical perspective on patient and public involvement, information on the patient and public experience is lacking [32]. In October 2019, a similar survey was distributed to 26 patients and family members who participated in recent guideline development (the 2014 Infection Prevention and Control committee to present committees). Eleven of the 26 patients and family members who were contacted responded to this survey (42% response rate). Eighty-two percent rated their overall experience on the guideline committee(s) as above average to excellent, with only one rating their experience as average and one rating their experience as very poor. Ninety-one percent of the respondents somewhat agreed or strongly agreed that the presence of someone with CF/caregiver/significant other improved the guidelines formation process, and all somewhat or strongly disagreed that the presence of someone with CF/caregiver/significant other inhibited discussion. Seventy-two percent somewhat or strongly agreed that the presence of someone with CF/caregiver/significant other improved the PICO questions chosen. Ninety-one percent somewhat or strongly agreed that the presence of someone with CF/caregiver/significant other improved the outcomes considered to be important.

When asked if they felt that the guideline benefited from their inclusion on the committee as a person with CF/caregiver/significant other, 10 of the 11 respondents felt the guideline benefited from their inclusion. The responses can be exemplified by one individual who stated, “[E]veryone brought a different view to the discussion and mine was not medical but that of a parent which very much plays a role in the care of the patient.” Another indicated, “I believe I was able to articulate unmet needs in current [CF] care that the … guideline could address.”

The individuals with CF and family members included on guidelines committees also keep the committee focused on the variation in experiences within the CF community. These members often remind the committee of the various choices in care and patient priorities, especially around advanced stages within the disease or during transplantation decisions. Their presence has ensured that the committee remembers the variety of care pathways available to targeted patients in the population and that these choices are considered when developing the PICO questions and recommendations. Textbox 1 presents a patient’s perspective on the guideline development process.

Textbox 1. A patient’s perspective.

The best medical care is a partnership between patients, families, and clinicians. As a patient, there are a few important ways we contribute to the guideline development process. While CF clinicians are no doubt experts in cystic fibrosis, we are the experts on where the “rubber meets the road” in CF care. Having a voice and a vote ensures that guidelines are feasible and more likely to be accepted by the wider CF community. When not directly participating on a specific committee, the opportunity to comment on guidelines is important as it still gives me a voice and input on the guidelines that will affect my care. Our experiences as patients or family members of someone with CF give us a unique viewpoint that often brings up symptoms or issues that might go unrecognized, and therefore left out of guidelines. Finally, there is the feeling of empowerment that comes with being treated as a colleague and not just a patient [33]. The role of guidelines continues to grow as evidence-based medicine becomes the standard. Those guidelines inform the care we receive and are expected to adhere to. Coproduction of guidelines is vital to ensuring that patient wishes and needs are always at the center of guideline development.

Conclusion

The inclusion of PPI in CFF guideline development since 2004 has dramatically strengthened the culture of the organization’s guideline development. With the advent of Community Voice, the CFF has been able to reach and partner with a broader range of individuals with CF and their family members, allowing more perspectives to be heard. Despite disease-specific challenges, the CFF has been able to utilize technology to incorporate multiple patient and family perspectives into guideline development. These voices also improve the way the CFF talks about and develops educational material related to the guidelines. Reviews of these materials by individuals with CF and family members ensure that the language used is understandable and culturally appropriate.

The surveys that were conducted show that clinicians, patients, and family members believe that the lived experience is an essential aspect in the creation of guidelines. This feedback has encouraged CFF to continue to explore additional ways to involve community members in guideline development and implementation.

The methods CFF uses to include patients and families in guideline development can be adapted to other conditions, both chronic and acute, as all patients and their family members can provide insight into their experiences with their conditions. While CFF has a highly connected and activated population mobilized through Community Voice, partnerships between clinicians, patients, and family members can be built and fostered in many different ways, including one-one relationships, quality improvement projects at the local level, and developing overarching care guidelines for an entire disease population.

Overall, the importance of patient and public involvement and partnership in CFF guideline development was summarized by one individual with CF/family member who stated in the survey that "[t]he personal insight on how things truly are from a CF patient or caregiver is invaluable."

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Conflicts of Interest

SHE, KF, and ETN report no conflicts of interest. WCH reports receiving honoraria from the Cystic Fibrosis Foundation for volunteer committee work unrelated to this manuscript.

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Abbreviations

CF: cystic fibrosis
CFF: Cystic Fibrosis Foundation
IOM: Institute of Medicine
GIN: Guidelines International Network
PPI: patient and public involvement

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Viewpoint

Guiding Pay-As-You-Live Health Insurance Models Toward Responsible Innovation in Health

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Abstract

While the transition toward digitalized health care and service delivery challenges many publicly and privately funded health systems, patients are already producing a phenomenal amount of data on their health and lifestyle through their personal use of mobile technologies. To extract value from such user-generated data, a new insurance model is emerging called Pay-As-You-Live (PAYL). This model differs from other insurance models by offering to support clients in the management of their health in a more interactive yet directive manner. Despite significant promises for clients, there are critical issues that remain unaddressed, especially as PAYL models can significantly disrupt current collective insurance models and question the social contract in so-called universal and public health systems. In this paper, we discuss the following issues of concern: the quantification of health-related behavior, the burden of proof of compliance, client data privacy, and the potential threat to health insurance models based on risk mutualization. We explore how more responsible health insurance models in the digital health era could be developed, particularly by drawing from the Responsible Innovation in Health framework.

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KEYWORDS

digital health; client; Pay-As-You-Live; quantified-self; social contract; health insurance; responsible innovation in health; health system; health data; health inequity; health determinants

Introduction

As health systems struggle to successfully implement a digital transition in care and service delivery [1], the ubiquity of mobile technologies combined with the emergence of the "quantified-self" movement has already generated a phenomenal amount of data on the health and lifestyle of individuals [2,3].

Pay-As-You-Live: When Digital Health Technologies Influence New Insurance Models

Because of important financial incentives in the health sector, generated in part by certain social and health policies [4], health insurers can now create a new insurance model called Pay-As-You-Live (PAYL), which extracts value from user-generated data [5].

Partly driven by the success of the quantified-self movement [5], PAYL is a variant of the Pay-As-You-Drive care insurance model, which relies upon a GPS device (embedded in the car or a mobile application) to track client driving behavior to inform insurer decisions regarding increases or decreases in insurance premiums [6]. The insurer sends comments, information, and suggestions in real-time (via SMS text messaging or email) to inform the client about the appropriate driving behavior to adopt [5]. In this way, the insurer identifies the risks and leaves it up to the insured to decide, via the quality of their driving, the amount they will pay for the insurance.

As an interactive form of insurance, the principle of the PAYL model is to support clients in the management of their health. With real-time feedback, many insurers offer clients mobile applications and sensors to motivate them to adopt healthy behaviors and good lifestyle habits [5,7]. When using these applications and sensors intended to collect data, clients must share a multitude of health data with the insurer (for instance, weight, heart rate, eating habits, calorie intake, sleeping habits, places frequented, blood pressure, and clinical history) to track progress and judge compliance with the insurer's recommendations. Clients who have achieved the defined objectives or have demonstrated a positive evolution over time are rewarded financially, especially in the form of bonuses (eg, capital of points) or reductions in the price of their insurance contract. As such, PAYL insurance models aim to reward clients who adopt healthy behaviors as much as they "punish" those who seem to choose to do otherwise.

Despite significant promises for clients, there are, in fact, critical issues that remain unaddressed, especially as PAYL models have the potential to significantly disrupt current insurance models and disproportionally impact more vulnerable segments of the population. In this paper, we discuss the following issues of concern: the quantification of health-related behavior, the burden of proof of compliance, client data privacy, and the potential threat to health insurance models based on risk mutualization. We then explore how more responsible health insurance models in the digital health era could be developed.

Challenges to Quantifying Complex Individual Behaviors

The quantification of behavior derives from the assumption that life can be converted into digital data, or that quantitative measures of different activities and behaviors are constitutive of the person [8]. The reduction of individuals to a set of standardized measurements and quantifications poses two major problems.

First, there is a risk of omitting the complexity that surrounds individual and collective behaviors, particularly by underestimating the impact of systemic social determinants of health [3,9]. For populations whose health problems stem from unmet socioeconomic needs, not only is their health impacted by poor housing, a lack of transportation or education, underserviced neighborhoods (eg, food deserts), or strenuous jobs, but they may also lack the means to adopt and record daily health behaviors predetermined by insurers [10]. For example, a person living in an underserviced neighborhood is less likely to have access to adequate sports facilities and, therefore, is less likely to be physically active enough to achieve the performance goals required to stay healthy as determined by insurers [10,11]. The same is true for a single parent who does not have access to childcare services. In addition, a person suffering from depression will find it difficult to be physically active or to prepare and eat healthy meals [12]. People in financially precarious situations also have greater difficulty buying nutritious and healthy food and having access to alternative food systems (eg, food swamps) [13,14]. Thus, vulnerable populations could face a double disadvantage if they are subjected to the PAYL model: They will face more expensive insurance policies as a result of systemic barriers that already affect their living conditions and health problems [15,16].

Second, while digital health technologies may potentially have an added value for people who are relatively young, healthy, educated, financially stable, and living in safe and serviced neighborhoods, demonstrating whether the PAYL model improves health outcomes remains challenging. Because individuals subjectively experience their life trajectory, their history, and their environment [17], some may become dependent on technology as they focus on achieving the required health goals, creating a sort of tyranny of self-measurement. Furthermore, individuals with low levels of digital and health literacy may have difficulty managing and interpreting large amounts of data and health information [9]. Such a situation could lead to other problems, including stress, isolation, low self-esteem, deterioration of quality of life, and cognitive overload ("fatigue alert") [18,19].

Placing the Burden of Proof on Clients

To prove that they are respecting the terms of their health insurance contracts, clients must demonstrate that they have adopted and followed through on a set of predetermined health behaviors; for example, they must exhibit that they have taken the required daily number of steps, eaten healthily, frequented green areas, or complied with their doctors' instructions. Taken on by the client, the burden of proof becomes problematic, especially when the basis for the insurance algorithm's determinations of the client's instructions and personalized objectives are unknown or ambiguous. Moreover, the insurer can interpret data in different ways. Indeed, it is difficult to know which criteria and parameters are retained by the insurer to estimate risks and calculate the insurance premium.

As an illustration, a person who trains regularly will have optimal physical health, at least theoretically. However, some physical activities are associated with a higher risk of accidents and injuries (eg, muscle and joint injuries, ligament rupture, osteoarthritis) [20-22]. How will the algorithm distinguish risk-avoidance behavior from risk-taking behavior in this case? Using user-generated data to neutralize risks, especially as certain physical damages become predictable, offers a major advantage to the insurer: both rewardable risk-avoidance behavior and risk-taking behavior may remain profitable for the insurer. Payment based on personal risks would, in the majority of cases, result in the client incurring all or a large part of the costs in the event of damage.

Extracting Value From Client Data

This issue of carrying the burden of proof also raises the question of how the value generated by the client's data will be shared. Data collection is a two-way process that involves continuous interaction between the client and the insurer [5,15]. Since data are collected on the basis of the individual's activities and behaviors, the client is a central contributor to the creation of value for the insurer. Thus, the generated value should be shared between the client and the insurer.

Client-generated data under a PAYL insurance contract could also be used for other purposes, whether or not the client has consented to these other purposes. On this point, regulatory requirements may vary and have different effects depending on the jurisdiction. Health data can be sold or outsourced to data brokers who specialize in selling medical forecasts to other third parties, such as employers, life insurers, and credit companies. Because the information asymmetry currently favors insurers, several data collection issues arise [5,15]. These include, for example, conformity with the country's sociopolitical norms and regulations; compliance with the principles of freedom and equal rights (eg, possibility to request the deletion of data, or the "right to be forgotten"); client consent to all data collection; client knowledge of data collected by insurers; and data use (or re-use) transparency.

Mutualization Versus Individualization of Risk

As a social value [23], the promotion of digital health can no longer be examined outside of the social contract that reflects every society's values [24]. With the PAYL model, publicly funded and universal health systems could gradually migrate from a model of collective solidarity, where risks are mutualized and where citizens receive services corresponding to their needs rather than their ability to pay, to a transactional model, based on the calculation of individual risks. Indeed, the high visibility enabled by this model when calculating individual risks will most likely impact the values of solidarity that characterize collective insurance models [5,15]. It can be hypothesized that not only will the most vulnerable groups be disproportionally affected, but the very basis upon which collective insurance models work will also be eroded.

Guiding Health Insurance Models Toward Responsible Innovation

The PAYL insurance model can be seen as an innovation, but one that has many pitfalls. To help guide the development of health insurance models that both leverage digital health technologies and meet population health needs, the Responsible Innovation in Health (RIH) framework developed by Silva et al [25] offers an interesting starting point. To our knowledge, the RIH framework is one of the few frameworks that go beyond innovation ethics and includes, in an integrated manner, organizational, environmental, health system, and population health aspects. Furthermore, it emphasizes a collective approach that can shed light on various private, public, and mixed health insurance models while keeping the effectiveness and safety of health innovation at the center of stakeholders' reflections and actions. This framework aims to support equitable and sustainable health care by fostering the development of innovations along 5 value domains that include 9 attributes [25,26]. Table 1 illustrates how the RIH value domains and attributes can be applied to set responsibility objectives for digitalized health insurance models.

The RIH framework invites those who develop insurance models to respond to the most pressing population needs while reducing health inequalities. To this end, stakeholders who have knowledge about and power over various determinants of health could be involved in identifying and defining a broader digital health dataset, one that makes explicit the systemic facilitators and barriers that affect how people live. This could provide a fairer and more valid representation of living conditions and, eventually, support personalized priorities and objectives according to the contextual factors affecting individual health behaviors. To better address health system needs, insurance algorithms could focus on helping clients manage chronic diseases or comorbidities that put a strain on health systems. An optimized, affordable, and easy-to-use platform or mobile application could be customizable according to clients' data plans (which can be very expensive in North America) and integrate digital and health literacy functionalities to enable clients to make the most out of their health data. A business model that provides value to both clients and society could share, following a clear data stewardship model, the collected health data with population health researchers who study, for instance, the impact of environmental factors on diabetes, heart disease, sleep disorders, etc. Finally, energy-intensive server farms could run on clean energy to reduce smog levels as well as the incidence of respiratory diseases associated with air pollution. In other words, the RIH framework offers a lens to reconsider how the mutualization of risks may spur innovative insurance models in which benefits may also be mutualized more broadly.

Table 1. The application of RIH value domains, attributes, and responsibility objectives for digitalized health insurance models.

Value domain and attribute	Objective
Population health value	
Health relevance	Respond to the collective needs of the population.
Ethical, legal, and social issues	Mitigate the ethical, legal, and social issues related to health data privacy and security.
Health inequalities	Contribute to reducing inequalities in access to health care and services (eg, digital and health literacy).
Health system value	
Inclusiveness	Work with stakeholders (eg, clients, health care providers, technology providers) throughout the innovation lifecycle.
Responsiveness	Appropriately respond to important health system needs (eg, service delivery gaps or demographic changes that call for alternative insurance packages).
Level and intensity of care	Examine at what level in the system appropriate care can be delivered safely and effectively.
Economic value	
Frugality	Provide an optimized, affordable, and easy-to-use solution to all clients.
Organizational value	
Business model	Follow a business model that provides value to both clients and society.
Environmental value	
Eco-responsibility	Reduce the environmental footprint caused by the production and use of digital technology and the development and use of digital infrastructures and algorithms (eg, servers).

More specifically, if we take the example of clients with low income and with type 2 diabetes, a responsible digitalized health insurance model could capture secure and private data on their living conditions (eg, type of work, types of services and amenities available in their neighborhoods, time constraints related to childcare duties, weekly budget), comorbidities (eg, retinopathy), and digital and health literacy (eg, mother tongue, reading and comprehension levels, knowledge on their disease). It could improve access to mobile technologies and data plans and appropriately secure clients' consent to share their anonymized health data for health research on type 2 diabetes. In this way, personalized health objectives and behaviors would take into consideration all diabetic clients' contextual facilitators and barriers. Furthermore, an application could help clients find weekly sales on fresh fruits and vegetables in their area, suggest a safe walking route to the store (if possible), recommend an easy recipe to cook products on sale, and provide a bit of information on how the purchased products help to keep them healthy. As such, rather than focusing on risk-taking or avoidance, healthy behaviors would be contextually scaffolded, and the insurance premium would reward the capturing and sharing of data for research purposes as well as a reduction in the level and intensity of care (eg, reduction in hospitalization rates).

Of course, to ensure the success of such a model, individuals must be willing to share a multitude of data that can provide a holistic picture of their context and needs. This is a major challenge, particularly given the lack of public confidence in how the data will be used in the future, especially around issues of confidentiality, privacy, commercial use, and discrimination [27]. It is essential to have transparent data governance structures and models to ensure the responsible and accountable use of the data for the benefit of the society. In order to increase and maintain confidence, it is also important to go beyond strict legal compliance and ensure real public involvement in data governance through information, transparency, and control [27].

Conclusions

With the PAYL model, health insurers seek to leverage new digital technologies and value extraction models while adapting to their clients' shifting health needs. However, as the latter increasingly includes diversified populations whose health is conditioned by broader determinants over which they have little, if any, control, this paper aims to shed light on problematic assumptions and the modus operandi behind this emerging model: the quantification of complex behaviors; shifting the burden of proof to clients who are, in a sense, presumed guilty unless digital trackers can show otherwise; the lack of transparency in how economic value is extracted from client data; and how such models undermine the very principle which, several decades ago, made health insurance a responsible public policy innovation—the mutualization of risk.

At the same time, the PAYL model remains an innovative approach that could help health systems become more efficient and equitable, particularly by supporting the development of healthy habits and adherence to care treatments. Yet, to achieve these objectives, we must consider the complexity inherent in the lives of individuals and communities, as well as the principles that define the social contract of our society, which includes the protection of vulnerable populations. In this respect, the RIH framework could be a major lever to guide the development of responsible digitalized health insurance models that adequately respond to and support the mission of health systems.

In conclusion, by analyzing the PAYL insurance model as a health innovation, this paper contributes to the scholarship by comprehensively applying the RIH framework to explore responsibility features that go beyond innovation ethics. In doing so, this paper also highlights the way in which an alternative digitalized health insurance model could be further developed by public or private institutions, thus providing a knowledge base for future studies.

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Authors' Contributions

HA and LR produced the first draft of this manuscript and received input from RRO, PL, SBMC, MS, MAAA, and JPF. All authors read and approved the final manuscript.

Conflicts of Interest

None declared.

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Abbreviations

PAYL: Pay-As-You-Live
RIH: Responsible Innovation in Health

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Original Paper

Engaging Youth in the Bipolar Youth Action Project: Community-Based Participatory Research

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Abstract

Background: We describe the methodological dimensions of community-based participatory research through a description of study design, youth engagement, and methods/processes in the cocreation of knowledge within a Canadian study, the Bipolar Youth Action Project. This collaborative partnership—carried out by a team composed of academic, community, and youth partners—was designed to investigate self-management and wellness strategies for young adults living with bipolar disorder.

Objective: The aim is to describe the opportunities and challenges of this collaboration and to reflect upon the process of involving youth with bipolar disorder in health research that concerns them, and share lessons learned.

Methods: The project was conducted in multiple phases over 2 years: (1) grant-writing, with youth contributing to the process; (2) recruitment, in which 12 youth were selected and trained to help shape and conduct two research forums; (3) the first research forum, where more youth were consulted about the strategies they apply to stay well (self-management strategies); (4) data analysis of Forum I findings; (5) research Forum II, which consulted youth with bipolar disorder about knowledge translation of Forum I findings; and (6) data analysis of Forum II findings. Youth peer researchers with bipolar disorder were involved in a significant capacity at every stage in the process.

Results: Of the initial 12 youth peer researchers, 7 remained on the project from the recruitment phase until the project ended. They collaborated in the creation of two youth research forums that consulted youth with bipolar disorder on their self-management strategies.

Conclusions: This article shares what was learned from the process of partnering with youth with bipolar disorder in a community-based participatory research study.

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KEYWORDS

community-based participatory research; bipolar disorder; adolescent; young adult; youth; participatory research

Introduction

Bipolar disorder is a type of mood disorder characterized by periods of depressed and elevated (manic or hypomanic) mood states, with corresponding changes in thinking and behavior [1]. The typical onset of bipolar disorder occurs in late adolescence to early adulthood [2]. According to the World Health Organization, the condition is the 6th leading cause of disability among people aged 0-59 years in higher-income countries, and 8th in lower-income countries [3]. Globally, bipolar disorder is the 4th greatest cause of disability-adjusted life years in people aged 10-24 years [4]. Youth, therefore, are a key target group for early intervention and support, particularly given that interventions for bipolar disorder may be more effective for younger adults than for their older counterparts [5,6].

Fostering self-management strategies—that is, the plans and routines that a person with bipolar disorder uses to promote health and quality of life [7]—is viewed as a key element of ensuring optimal health in people with the condition. A solid body of evidence now exists on self-management in adults with bipolar disorder [7-11]. However, much less is known about effective self-management for bipolar disorder in youth populations [10].

Community-Based Participatory Research

The research methods of the Bipolar Youth Action Project were guided by the Collaborative RESearch Team to study psychosocial issues in Bipolar Disorder (CREST.BD) [12], a Canada-based network dedicated to collaborative research and knowledge translation in bipolar disorder. The CREST.BD network specializes in community-based participatory research, a participatory research approach in which academic researchers and community members work in partnership [13]. Community-based participatory research aims to shape research around community priorities, emphasizing knowledge generation that contributes to the community and social change [13-15]. Community-based participatory research can be viewed as a "philosophy of engagement" [16] rather than a discrete method per se; instead, diverse methodological approaches can be applied within the frameworks of the research method. The CREST.BD team, who are informed by a decade of research and integrated knowledge translation, developed a specific model of community-based participatory research for bipolar disorder [17]. The model builds on the strengths of the community of people with bipolar disorder and has been successfully applied across diverse projects in populations of adults with bipolar disorder [9,17-19].

A logical progression in CREST.BD's program of research was to explore the application of community-based participatory research approaches in youth and young adults living with bipolar disorder. Community-based participatory research has long been viewed as an effective approach for working with underserved populations, gaining increasing support for its use over the past twenty years [20-23]. Seldom heard populations, including populations of youth [24], voice a need for innovative approaches to address issues of social, contextual, language, and cultural factors faced in mental health treatment systems. For youth facing mental health challenges, community-based participatory research approaches hold potential to amplify well-being and personal strengths [25], increase access to mental health services and information [26], and enhance methodological rigor and implementation of research findings [20,24].

Diverse and international youth populations have now been engaged in community-based participatory research projects [24]. In the mental health arena, these approaches have been used to advance knowledge on substance use in youth [27]; suicidality in American Indigenous youth [21]; trauma, psychiatric issues and educational and behavioral outcomes in Cambodian American youth [28], and bullying in elementary-aged youth [29]. The potential assets of community-based participatory research approaches in youth mental health research are clear. However, there are particular challenges in participatory research with youth, including difficulties concerning power differentials [30], problems maintaining engagement [31], concerns of disclosure and anonymity [32], and differing research goals between academic and peer research teams [30].

In summary, we know that self-management strategies are critical for health and quality of life in people with bipolar disorder, but a gap exists in self-management research specifically addressing youth with the condition. A specific model for community-based participatory research in adults with bipolar disorder has been developed, but little is yet known about the application of this approach in youth. This article traces the lifespan of a 2-year community-based participatory research-informed study engaging youth with bipolar disorder as peer researchers. We describe how youths' roles as peer researchers presented both opportunities and challenges to more traditional research and knowledge translation activities. We also outline the lessons learned from this process.

Methods

Project Aims and Overarching Design

The Bipolar Youth Action Project was a 2-year youth-driven research project with a primary aim of building knowledge on (1) effective self-management strategies for youth with bipolar disorder, and (2) preferred knowledge translation methods for sharing this knowledge with other youth with bipolar disorder and their supporters. A secondary aim was to develop the knowledge base on the application of community-based participatory research in youth living with bipolar disorder. Youth with bipolar disorder were integral to each stage of the Bipolar Youth Action Project, as funding coapplicants, peer researchers, research participants, and knowledge brokers. Previous Bipolar Youth Action Project publications have described the project's specific methods [33] and provided a qualitative analysis of youth preferences for knowledge translation methods for online health information [34]. Here, we take a deeper dive into our secondary aim, which is to describe and share reflections on the community-based participatory research approaches we undertook in the Bipolar Youth Action Project. In the following sections, we describe the specific methods we undertook across the various study phases.

Prefunding Phase

The Bipolar Youth Action Project was funded by the Vancouver Foundation, a British Columbia-based funding agency dedicated to supporting community-focused research. The funding application was coproduced in equal partnership by two organizations: the Bipolar Disorder Society of British Columbia (BDSBC; now Vancouver BC-based Stigma-Free Society), an organization providing social support and services for people living with bipolar disorder based in Victoria, British Columbia, and CREST.BD, headquartered in Vancouver. In order to generate pilot data for the funding application, the Executive Director of BDSBC conducted a focus group with five youth with bipolar disorder who were associated with the BDSBC.

The youths were recruited through announcements at BDSBC's support groups and via advertisements within the community and in a local newspaper. Youth applicants were required to submit a resume and cover letter and were interviewed by the Executive Director of the BDSBC and by the project's Principal Investigator. The inclusion criterion for eligibility was self-report of a health care provider diagnosis of bipolar disorder (type I, II, or not otherwise specified). Two of the youth who participated in the focus group self-selected to serve as coapplicants on the funding application and collaborated on the identification of project methods.

Phase 1: Team Establishment and Capacity Building

Team Establishment

Central to the Bipolar Youth Action Project was the establishment of the Youth Action Group, a group of young adults aged 20 to 25 years living with bipolar disorder, who served as peer researchers for the duration of the project. The BDSBC recruited 12 youth by advertising the project within their community network and interviewing prospective members, with the expectation that there would be some attrition over the study's 2-year span. The recruitment process and inclusion criteria were the same as in the prefunding phase, with the addendum that members of the Youth Action Group must also be able to complete a 2.5-hour research ethics course (the Tri-Council Policy Statement Course on Research Ethics). Two “Co-Leads” self-selected to take on the additional responsibilities of governance and leadership of the Youth Action Group. The wider project team consisted of an academic researcher specializing in community-based participatory research in bipolar disorder, the Executive Director of the BDSBC, a specialist from an organization dedicated to youth engagement, health care providers (two psychiatrists who were coapplicants on the funding application and one mental health counselor), and a research coordinator.

Capacity Building

A series of four foundational training sessions were conducted with Youth Action Group members to build capacity. The sessions were focused on: the principles and implementation of community-based participatory research; qualitative, quantitative and graphic facilitation methods; research ethics and knowledge translation; and providing a grounding in CREST.BD's previous research exploring self-management in adults with bipolar disorder (see Table 1).

Graphic facilitation, or graphic recording, is a process of illustrating themes and ideas shared during discussions using a combination of text and imagery, typically on a whiteboard or large sheet of paper [35]. The rationale for training Youth Action Group members in graphic facilitation was twofold. First, it was a means of knowledge translation of findings from CREST.BD's research into adult self-management, as group members were tasked with visualizing previous findings as the academic team presented them. Second, introducing graphic facilitation to group members provided training in a knowledge translation method that could be utilized at the research forums.

Table 1. Phase I research training events.

Training Day	Purpose	Description
Research training day	YAG[a] team-building, education about CBPR, qualitative methods	Full day in-person event
Graphic facilitation day	Educate YAG about self-management and graphic facilitation methods	Full day in-person event
TCPS2 CORE course[b]	Train all YAG members in research ethics	Online research ethics course for researchers and research staff
CBPR[c] webinar	Refresh YAG knowledge of CBPR and qualitative research methods	Web-based presentation at the end of Phase 1

[a] YAG: Youth Action Group.

[b] The Tri-Council Policy Statement Tutorial Course in Research Ethics (Government of Canada Panel on Research Ethics, 2016).

[c] CBPR: community-based participatory research.

Phase 2: Forum I

Once the training phase was complete, team members co-designed and delivered the first of two “Youth Research Forums,” hosted at an event center chosen by the Youth Action Group. The primary research goal of Forum I was to yield new knowledge on self-management of bipolar disorder in youth. A secondary aim, determined by the Youth Action Group, was to share knowledge of bipolar disorder self-management and stigma in the form of group-designed workshops and presentations. In order to include the perspectives of youth in high school, participants in Forum I could be aged 16-25 years, and self-identified as living with bipolar disorder I, II, or not otherwise specified. Recruitment was undertaken jointly by the BDSBC, who reached out within its network, and Youth Action Group members, who distributed posters and volunteered at mental health awareness events.

At the opening of the day, Youth Action Group members delivered presentations and workshops to forum attendees, followed by a group-led mindfulness activity. Afterward, academic research team members with experience in qualitative methods moderated four 90-minute focus groups to discuss self-management strategies used by youth to stay well. Focus groups were digitally recorded, with Youth Action Group members acting as notetakers. Throughout Forum I, a graphic facilitation specialist visualized themes from the Youth Action Group presentations on a large sheet of paper hung on the wall (Figure 1).

A private area was available for participants to retreat to in case of distress, and health care providers (one female, one male) were available on-site to provide support as needed. One forum participant opted to bring a parental supporter with them to the event (but the supporter did not participate in data collection or discussions).

Figure 1. Erin Stewart Elliott, the project's graphic facilitator, presents her illustrations of the day's themes at Forum I.

Phase 3: Forum I Analysis

A Youth Action Group member transcribed the focus group audio recordings verbatim and ensured all identifying information was removed. A coding framework was collaboratively developed with the Youth Action Group member and two academic researchers; the Youth Action Group member also participated in the thematic analysis (findings reported elsewhere) [33]. The same member created a summary of findings to present to the rest of the group during Forum II planning.

Phase 4: Forum II

The primary goal of Forum II was to share the results of Forum I with participants and to consult on suggestions for knowledge translation and dissemination of Forum I findings. Inclusion criteria for Forum II were the same; both prior Forum I participants and new youth participants were welcome to join. Youth Action Group members shared the results of Forum I using presentations, arts-based methods, and group activities. As at the previous Forum, a graphic facilitator illustrated themes discussed and shared throughout the day on a large wall-mounted sheet of paper.

After the presentations and workshops, youth participated in a World Café to discuss avenues for knowledge translation. World Café is a method of structured conversation in which participants move between multiple tables, each with a designated topic of discussion. Academic researchers served as facilitators at each of three tables, and one table was facilitated by the Youth Action Group Co-Leads. Other members joined discussions as participants. The conversations at each table were recorded.

Phase 5: Forum II Analysis

World Café audio recordings were analyzed by a Youth Action Group member and another researcher. Detailed descriptive notes were written, eliminating identifying information, with tallies tracked of concrete suggestions made for knowledge translation.

Ethical Considerations

The University of British Columbia Behavioural Research Ethics Board and the Island Health Research Ethics Board granted ethics approval for the Bipolar Youth Action Project. Youth aged 19 years or above provided written consent to participate in the study; youth aged 16 to 18 years provided both their written assent and written consent of a parent or guardian. Consent packages were written in plain language to a Flesch-Kincaid reading level of grade 8 and included discussion around the permanency of media (photos and video) produced as products of the study. For data-gathering, consent to record audio was a prerequisite for participation in both forums, but participants, including the Youth Action Group, could decline consent for photography and videotaping.

Compensation Considerations

Youth Action Group members were compensated at a rate of USD42 (CAD50) per monthly meeting; Co-Leads were compensated at a rate of USD55 (CAD65) per meeting. One member was employed on contract to conduct the transcription, and support the coding and data analysis, of Forum I results, and the transcription and initial analysis of Forum II results. Youth Action Group members received reimbursement for childcare and travel expenses, and meals were provided at all meetings.

Results

Recruitment and Retention

At the outset of the Bipolar Youth Action Project, 12 young adults (aged 20-25; 10 females and 2 males) living with bipolar disorder were recruited to the Youth Action Group. All members were Caucasian and resided within urban areas. One group member was a parent. At the end of the 2-year project, 7 of the 12 original members (58.3%; 6 females and 1 male) remained engaged, a notable retention rate for a youth project of this length. Three members of the Youth Action Group (all female) discontinued participation in the because of illness relapse or reported difficulties balancing project responsibilities with university and work commitments. 2 additional youth (1 female, 1 male) discontinued without citing a reason, and the team was unable to reach them for follow-up. Of note, all attrition of Youth Action Group members was before the final training session; from this point until the project end, the seven members remained engaged.

Youth Roles and Capacity-Building

Youth Action Group members' participation in the Bipolar Youth Action Project was hybrid in nature. First, the members acted as paid peer researchers. Their role included helping to inform and guide the research direction, organize and present at the research forums, and disseminate study results. The members attended monthly meetings, participated in training sessions, and maintained continuous communication with the academic team via email, and with one another via Facebook. Throughout the project, members took on project roles and built capacity in areas that interested them, including meeting agenda cocreation (n=2); event planning (n=7); social media management and outreach (n=4); creation of infographics and video presentations (n=7); public speaking (n=7); data analysis (n=1); event cohosting (n=4); and knowledge translation of self-management strategies into engaging presentations (n=7).

Second, Youth Action Group members acted as research participants themselves. At Forum II, the group explicitly contributed knowledge about how best to share youth bipolar disorder self-management strategies with the wider community, and their experiences with participating in the project as peer researchers was an important source of knowledge about youth community-based participatory research engagement.

Facilitating Youth Action Group members' contributions and roles throughout the project required a flexible approach, with the community partner and academic team adjusting their work schedules to accommodate work and school schedules. Attention was paid to considering how to develop rapport effectively, convey information in accessible and engaging terms, assign appropriate quantities of work, and aid the youth peer researchers in taking ownership of the project. The group members worked most effectively with clear deadlines and consistent email communications.

Training events contributed to cohesion and trust-building within the overall team, which helped to model, build, and maintain effective working relationships and retain participants. Although significant staff time and explicit funding were set aside for Youth Action Group training during the eight months of phase I, in practice, training was ongoing throughout the project on a less formal basis. Given the length of the study, the members would likely have benefitted from refresher training on concepts learned earlier in the project.

Youth became more secure in their roles and in taking ownership of the Bipolar Youth Action Project as it progressed, demonstrating a greater willingness to take on leadership roles after they had been delineated or modeled by the community partner and academic researchers. For Forum I, five of the seven Youth Action Group members elected to give workshops on specific topics related to mental health. During this process, the academic team supported the members in selecting topics and encouraging them to develop their ideas. The chosen topics were (1) living with mental health stigma, (2) creating a personal mindfulness toolbox, and (3) "Leading Extraordinary Lives" (Table 2). Although the members had been trained in graphic facilitation, they chose not to partake in this at Forum I.

Youth Action Group members were much more confident assuming leadership for the second Forum and wrote and delivered workshops with minimal involvement of the academic researchers. Of the 7 Youth Action Group members, six were split into pairs, and each chose to present on themes from Forum I. The chosen topics were: (1) health and fitness, (2) in-the-moment strategies for managing shifts in mood, and (3) developing support networks (Table 2). As with Forum I, Youth Action Group members demonstrated creativity and passion towards the topics chosen; however, they were notably more confident in the development and execution of their ideas for Forum II and required less input and encouragement from the academic team. At Forum II, Youth Action Group members again elected not to partake in graphic facilitation of presentations and workshops, although they did create some writings and illustrations with other participants during the introduction phase of the day.

Table 2. Youth Action Group–led and designed workshops.

Workshop topic	Description
Forum I	
Stigma	One member and the community partner delivered a presentation explaining the stigma of bipolar disorder and how to live with it.
Mindfulness	Two members introduced the topic of mindfulness and led participants in creating personal mindfulness toolboxes.
Leading Extraordinary Lives	Two members presented their personal health stories, with the underlying message that it is possible to live an extraordinary life with bipolar disorder.
Forum II	
Health and fitness	Two members shared their personal health stories as a means of knowledge translation of Forum I findings of self-management through diet and fitness.
In-the-moment strategies	Two members used props to demonstrate strategies for self-management while amid a depressive or manic/hypomanic episode, as a means of knowledge translation of Forum I findings.

Evolving Communication

A common workflow was established reflexively over time, based on group feedback and the academic team's observations of how engagement varied as a function of project stage, volume of communication, and level of structure provided by the academic team. Email communications served as an important supplement to monthly in-person meetings (especially as the project was being conducted from Vancouver Island and the city of Vancouver), and the academic research team noted that the greater the number of email communications, the more engaged the Youth Action Group appeared to be. Often, deadlines would be agreed upon during in-person meetings, but friendly check-ins during the monthly helped members to meet these deadlines and to be open about any roadblocks or challenges they faced.

Data Analysis

As noted elsewhere [33], Youth Action Group members expressed that conveying the forum findings in peer-reviewed papers was a priority for them. One member stated an interest in becoming more heavily involved in data collection and analysis. This individual was hired as a contract research assistant to transcribe the Forum I focus group proceedings, and then to take the lead as a peer researcher in the analysis of the focus group findings. The process of involving this peer researcher was mutually beneficial. It helped to develop their capacity in their area of interest, and their lived experience lens was valuable in selecting themes likely to hold relevance towards youth living with bipolar disorder.

Discussion

Maintaining Engagement

A particular challenge in engaging youth as coproducers of health research is accommodating their fluctuating school and work schedules [31,36]. In the case of the Bipolar Youth Action Project, it was also expected that Youth Action Group members' active mood episodes could affect retention and participation. Indeed, for these reasons, some group members did leave the project during Phase 1; however, participation remained consistent after that.

The following are several recommendations to maintain youth engagement.

Foster Motivation With Open Communication Around Project Goals and Codevelop Goals to Include Participation of All Team Members

The academic research team found it helpful to ground research activities in the context of a grander purpose. It was important to ensure that youth peer researchers understood how specific activities related to the project aims; research activities that seem obvious to academic team members can appear to be nonsequiturs to youth if they are not informed of the reasoning behind them. Concrete actions that the academic team took towards this aim included: creating an open atmosphere where questioning was encouraged; conveying information through means that involved active and creative participation of the peer researchers, such as graphic facilitation; hosting a final training session that re-stated learnings from previous sessions; opening discussions for youth expertise, and maintaining consistent communication.

Reassure Youth That Their Input Is Valued and Take Care to Facilitate Their Input

Peer researchers may feel discouraged from contributing to discussions if they feel their input is not valued [37]. Therefore, creating an environment in which peer researchers' thoughts and opinions are respected helps them to feel their input is legitimate [38]. Community partners can be useful towards this aim, serving as a liaison between academic and peer researchers, and representing the interests of the target population [37]. The familiarity and trust Youth Action Group members felt with the community partner from the project outset aided them in expressing their points of view. In addition, cocreation of meeting agendas with Youth Action Group Co-Leads ensured that youth input would be woven directly into the structure of discussions.

Assign Concrete Responsibilities to Serve as a Mechanism for Engagement

In the context of youth-adult partnerships in mental health research, a balance of flexibility with clear expectations has been found beneficial [39]. Indeed, Youth Action Group member engagement peaked before and during research forums, when tasks to be executed were most concrete. Throughout more abstract phases of the Bipolar Youth Action Project, youth peer researchers were less engaged and required more follow-up to ensure they were aware of meeting times and tasks that needed completion. At these times, group members may have felt unsure about what they were meant to do and had had little reason to think of the Bipolar Youth Action Project amid competing priorities. Establishing more concrete responsibilities early in the project, and contextualizing them as vital for the forums, would likely have helped the youth become more engaged in the project in its earlier stages.

Prioritize Effective Communication

Disengagement in participatory research has been attributed in part to mismanagement of roles and a lack of clear, open communication [40]. Keeping consistent contact, such as meeting reminders and communication about assigned work, can improve output and engagement [41]. Throughout the Bipolar Youth Action Project, consistent email communication enhanced peer researcher engagement. It was also important to convey information to members in terms that were accessible and considerate of their condition. The sharing of prior research findings during a training event using graphic facilitation methods provided grounding in CREST.BD's earlier work in bipolar disorder self-management in an engaging and self-relevant manner.

In CREST.BD's previous work, the pace of research has, at times, been seen as slow and frustrating by peer researchers [42]. A strategy of pre-emptive sharing of "research snapshots" with the group before extensive analysis of the data has been effective in past community-based participatory research projects conducted by CREST.BD [42]. This strategy was utilized between the first and second forum, at meetings, and through email communications. Early iterations of papers were shared with Youth Action Group members before submission, both for their input and approval, as well as to assure them that their efforts were seeing fruition.

Capacity-Building Over Time

Leadership

In CREST.BD's prior research, tensions have been experienced in terms of meeting projects' funded research goals in addition to peer researcher or community goals [42]. In the Bipolar Youth Action Project, Youth Action Group members tended to focus on goals relating to advocacy and immediate action, whereas the academic researchers were oriented towards research and knowledge translation goals. Negotiating these diverse orientations required a reflexive process of establishing spheres of leadership, supporting Bipolar Youth Action Project's peer researchers to put project outputs into practice and make an immediate impact during forums. At the same time, academic research team members focused more on enacting research goals. A recent publication in youth participatory research posits that, rather than exercising paternalistic direction over youth peer researchers in the interest of promoting research goals, it can be beneficial to consider youth goals "in parallel" to those of academic researchers: distinct, yet proceeding in the same direction [30].

Power Inequities

Unequal power differentials are a perennial challenge within participatory research [20,26,30,43], with academic researchers holding an advantage of greater scholarly knowledge, research experience, and status within research projects [44]. Peer researchers hold power and expertise in their own right, through lived experience [39,45], but this power may not be broadly acknowledged [44]. In projects like the Bipolar Youth Action Project, with younger peer researchers, these power inequities may be amplified. Youth, accustomed to hierarchical contexts at school and work, may find it unnatural to be called upon to collaborate as peers, presenting academic researchers with the task of providing necessary supports to encourage collaboration. Affording Youth Action Group members an arena in which to ask questions, and to research their ideas in a working context, was therefore viewed as an important opportunity for helping them to build capacity.

Time Considerations

Commentators in participatory research have cautioned that inadequate time for involvement by peer researchers can render their participation superficial. Researchers advocate for thoughtful training with time built in for flexibility and delays [40,46]. The "publish or perish" mentality can compel academic researchers to leave insufficient time for the unfolding of participatory research processes, and this risks rendering the involvement of community members tokenistic [40]. The 2-year timeline of the Bipolar Youth Action Project provided the academic team adequate time to reflect upon and adjust and communicate expectations of Youth Action Group members and allowed the members to build capacity over time. In working with youth, who may feel less comfortable assuming responsibility and stating their perspectives openly, longer timelines can provide ample space for the gradual building of capacity and confidence and assumption of responsibility.

Settle Collectively on a Workflow That Suits Both Parties and Provide Adequate Structure for Youth Contributions.

Establishing principles of work is foundational to creating positive working relationships within a peer research group [47], which in turn can equalize and enhance patient-led research [45]. The timeline of the Bipolar Youth Action Project allowed consideration towards developing a common workflow that suited the needs of the Youth Action Group. When group members lacked clarity, they were not confident in taking action. Therefore, the process that emerged was one of continuous communication between meetings through email, frequent check-ins, specified deliverables, and clear deadlines. The authors encourage academic researchers undertaking community-based participatory research with populations of youth to determine a workflow that suits youth peer researchers collectively and to be willing to commit extra time and resources to form structure, communication, and scaffolding to foster youth involvement.

Compensate Peer Researchers for Their Time and Effort

Within participatory research, failing to provide adequate compensation can lead to disengagement and disempowerment of peer researchers [40]. Compensation creates an environment of reciprocity, in which all participants feel valued [38]. Compensation takes multiple forms, including payment for work, reimbursement for expenses such as transit, provision of meals and snacks, demonstration of respect and appreciation, and public acknowledgment of contributions [38]. Within the Bipolar Youth Action Project, sharing meals at each meeting helped to build trust and mutuality between peer researchers and academic researchers. Monetary compensation reinforced member expertise in the Youth Action Group and demonstrated the value of their contributions.

Be Sensitive to Ethical Concerns

In the application of community-based participatory research with seldom-heard populations of youth, as in the Bipolar Youth Action Project, a considered approach to ethics is necessary to ensure safety [47]. Most evident is the need to ensure that health professionals are available should distress occur, and to carefully convey sensitive information [40,47].

Disclosure and Anonymity

A particular challenge within the Bipolar Youth Action Project was disclosure. In recent years, research approaches have shifted from championing anonymity towards the notion that it can be empowering for participants to choose for themselves whether they would like to remain anonymous [32]. However, bipolar disorder is undeniably stigmatized [48], and this presents a concern for many people with the condition [17]. Most group members were comfortable having their names and photographs shared as a part of the coproduction of materials, but some were not. It was challenging to simultaneously meet the goals of coproduction and empowerment within community-based participatory research with the need to protect confidentiality and when requested, anonymity. In the Bipolar Youth Action Project, this was resolved by only photographing and sharing the names of those who consented to do so, and through ongoing dialogues regarding when and where the Youth Action Group members were comfortable being identified. Of the many materials produced throughout the 2-year project, it was sometimes possible to include all members of the team while omitting their names and or diagnoses from the shared material. Continuous communication with Youth Action Group members was essential to ensuring that they felt empowered and credited when desired, without sacrificing confidentiality and anonymity of other members.

Relationships Between Community Organizers and Peer Researchers

Another ethical consideration underscored by this project was the reliance of the participants, often members of stigmatized, seldom heard, and at-risk populations, upon the services provided by research or community organizations involved in a community-based participatory research project. In the case of the Bipolar Youth Action Project, the majority of Youth Action Group members were recruited through their attendance at BDSBC support groups, which they may have depended upon for support and connection to others with bipolar disorder. This reliance on organizations involved in community-based participatory research may represent a form of implicit coercion, in that it could compel participants to remain involved not for the sake of the project, but for the sake of maintaining positive relationships with those providing essential health services to them. With this in mind, we recommend continually reassuring community-based participatory research participants that their relationship to these organizations and services is not contingent on their continued involvement, and that, regardless of the outcome of their participation in the study, the team's highest priority is that community members are not facing barriers to service access.

Limitations

There were some limitations to the Bipolar Youth Action Project. All Youth Action Group members were white and resided within an urban area. The study was conducted on the traditional territories of the Songhees and Esquimalt First Nations; however, no participants self-identified as having First Nations heritage, and we were unable to engage with local First Nations as a part of this project to gauge their interest in the research. Only binary gender identities were represented. Only 2 of 12 members were male-identified at study outset; at its conclusion, only 1 of the male-identified members remained. It is unknown if any LGBTQ+ representation was present in the group, as this information was not requested or shared. Our results are not, therefore, easily generalizable to racialized populations, populations with diverse gender and sexual identities, and populations living outside of urban areas.

Conclusions

This article has described challenges and lessons learned during a community-based participatory research project involving youth with bipolar disorder. As compared with other forms of participatory research, research that involves youth may require additional time, communication, support, and attentiveness to power differentials. Youth motivations may differ from adult and academic researchers, and it is important to codeliver on both youth-identified and academic research priorities. We hope that this article will contribute to the knowledge base on conducting participatory mental health research and aid others in the design of mutually beneficial participatory research projects with youth populations.

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Conflicts of Interest

None declared.

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Abbreviations

BDSBC: Bipolar Disorder Society of British Columbia
CREST.BD: Collaborative RESearch Team to study psychosocial issues in BD

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Original Paper

Supporting the Implementation of Connected Care Technologies in the Veterans Health Administration: Cross-Sectional Survey Findings from the Veterans Engagement with Technology Collaborative (VET-C) Cohort

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Abstract

Background: Widespread adoption, use, and integration of patient-facing technologies into the workflow of health care systems has been slow, thus limiting the realization of their potential. A growing body of work has focused on how best to promote adoption and use of these technologies and measure their impacts on processes of care and outcomes. This body of work currently suffers from limitations (eg, cross-sectional analyses, limited patient-generated data linked with clinical records) and would benefit from institutional infrastructure to enhance available data and integrate the voice of the patient into implementation and evaluation efforts.

Objective: The Veterans Health Administration (VHA) has launched an initiative called the Veterans Engagement with Technology Collaborative cohort to directly address these challenges. This paper reports the process by which the cohort was developed and describes the baseline data being collected from cohort members. The overarching goal of the Veterans Engagement with Technology Collaborative cohort is to directly engage veterans in the evaluation of new VHA patient-facing technologies and in so doing, to create new infrastructure to support related quality improvement and evaluation activities.

Methods: Inclusion criteria for veterans to be eligible for membership in the cohort included being an active user of VHA health care services, having a mobile phone, and being an established user of existing VHA patient-facing technologies as represented by use of the secure messaging feature of VHA's patient portal. Between 2017 and 2018, we recruited veterans who met these criteria and administered a survey to them over the telephone.

Results: The majority of participants (N=2727) were male (2268/2727, 83.2%), White (2226/2727, 81.6%), living in their own apartment or house (2519/2696, 93.4%), and had completed some college (1176/2701, 43.5%) or an advanced degree (1178/2701, 43.6%). Cohort members were 59.9 years old, on average. The majority self-reported their health status as being good (1055/2725, 38.7%) or very good (524/2725, 19.2%). Most cohort members owned a personal computer (2609/2725, 95.7%), tablet computer (1616/2716, 59.5%), and/or smartphone (2438/2722, 89.6%).

Conclusions: The Veterans Engagement with Technology Collaborative cohort is an example of a VHA learning health care system initiative designed to support the data-driven implementation of patient-facing technologies into practice and measurement of their impacts. With this initiative, VHA is building capacity for future, rapid, rigorous evaluation and quality improvement efforts to enhance understanding of the adoption, use, and impact of patient-facing technologies.

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KEYWORDS

eHealth; mobile health; patient engagement; telehealth; veterans

Introduction

Background

Health care systems are facing an era of unprecedented growth in the number of patient-facing eHealth technologies available. Personal health record portals, mobile health (mHealth) apps, clinical videoconferencing platforms, automated texting systems, and other such tools have great potential to reach, engage, and empower patients, to support access to and delivery of care, and to improve outcomes; however, this potential has not yet been widely realized [1-4]. The experiences of many health care organizations underscore that promoting patient adoption and use of these technologies is difficult, and their effective integration into routine care can be elusive. Different patients may have different levels of interest in using eHealth technologies and may choose to abandon use if their expectations are not sufficiently met [5]. Because these technologies have, in many cases, not yet attained widespread recognition or endorsement among health care providers, patient awareness of their availability may be limited [6]. Similarly, the extent to which patient-facing eHealth technologies fit into a patient's daily life and their larger technological milieu can also directly affect their perceived usefulness and benefits.

Recognizing the importance of these issues, a growing body of work has focused on how best to bolster patient adoption and use of eHealth technologies, monitor their use, and measure their impacts on processes of care and outcomes [2,7,8]. This work, however, has also faced challenges. Particularly, studies that have relied on secondary data (eg, hospital administrative data or technology activity log data) to measure exposure to and use of patient-facing technologies and their effect on health care utilization and outcomes have encountered other issues, the most problematic perhaps being missing data. Many covariates related to technology use (eg, health literacy, education level, income level) are simply not available through such data sources. However, these covariates are essential to understanding technology adoption and adjusting for confounding factors when modeling associations with outcomes. A similar challenge exists for outcomes of interest to health care organizations, including patient-reported outcomes and perceptions of care. These challenges speak to how important it is for health care organizations to consider developing infrastructure capable of offering more complete data and placing the voice of the patient in the foreground as the key stakeholder in efforts to implement and evaluate patient-facing eHealth technologies [2].

Transforming Care Through eHealth Technologies in the Veterans Health Administration

The Veterans Health Administration (VHA) has been a pioneering, national leader in developing patient-facing eHealth technologies and integrating their use into clinical practice. Similar to other health care organizations, the VHA has developed a range of such technologies intended for use by the patient population that they serve—veterans of the US military. Table 1 presents key categories of these technologies and a description of each.

Table 1. Select VHA patient-facing eHealth technologies [9].

Technologies	Description
Key telehealth services	
Remote patient monitoring	Health data (eg, blood pressure, weight, glucose level) is gathered by a device issued to the patient and that data is, in turn, sent to the patient's care team.
Video appointments	Camera and audio on smartphones, computers, tablets, and other devices are used to support a video appointment between a patient and their care team from the comfort of the patient's home.
Personal health record portal	VHA's[a] tethered patient portal offers health education resources and features that support transactions with the health care system (eg, medication refilling), communication between a patient and their VHA clinical team members (eg, asynchronous secure messaging), self-management support (eg, tracking behaviors and symptoms), and access to the content of the patient's medical record (eg, Blue Button).
Automated text-messaging	VHA's protocol-driven automated text-messaging system provides tailored support for condition-specific self-management and other health behaviors through one-way and two-way messaging.
Mobile apps	VHA has developed a suite of mobile apps (which are designed to address the unique needs of the Veteran population) intended to promote wellness and healthy behaviors, provide condition-specific self-management support, support other transactions with the health care system, and enhance clinical management.

[a] VHA: Veterans Health Administration.

In addition to those presented in Table 1, new VHA patient-facing technologies are in continual development. As these technologies have been developed and rolled out at different times and are intended to meet different needs, the extent of their adoption within the veteran population varies considerably [10-12]. The work of developing and implementing these technologies is the responsibility of VHA's Office of Connected Care, which oversees VHA's digital health strategy and is focused on improving VHA care through technology that engages veterans beyond traditional health care visits.

Recognizing the need for further insights to advance the implementation and evaluation of their portfolio of technologies, in 2016, the VHA Office of Connected Care, in conjunction with the VHA Quality Enhancement Research Initiative Program, funded an effort to recruit a group of veterans willing to make a long-term commitment to providing feedback on the latest VHA patient-facing technologies and helping the VHA understand their potential benefits. Branded the Veterans Engagement with Technology Collaborative, the overarching goal of this cohort is to directly engage veterans in the evaluation of new VHA patient-facing technologies intended to improve access to care, enhance care coordination, and support self-management, and in so doing, to create new infrastructure to support related research and evaluation activities. Soliciting the expertise of patients and integrating their perspectives into VHA's technology evaluation efforts demonstrates the values of participatory medicine, a patient-centered philosophy, in action. It also aligns with broader initiatives by VHA and its leadership to enhance veteran engagement in an effort to realize health care system improvements that resonate with the veteran population.

Importantly, the development and use of the Veterans Engagement with Technology Collaborative cohort was designed to meet the criteria for quality improvement and was subsequently reviewed by the Institutional Review Board at the Edith Nourse Rogers Memorial Veterans Hospital in Bedford, Massachusetts, and determined to be such [13]. This designation is an important step toward realizing the National Academy of Medicine's vision for learning health systems, where "science, informatics, incentives, and culture are aligned for continuous improvement and innovation, with best practices seamlessly embedded in the delivery process [14]."

Objectives

The specific objectives of the Veterans Engagement with Technology Collaborative cohort were to (1) identify the extent of exposure and use, as well as patient perceptions, of select VHA patient-facing eHealth technologies, (2) understand characteristics and determinants associated with adoption and use of specific VHA patient-facing eHealth technologies, and (3) examine the impact of patient-facing eHealth technology use on select patient-reported outcomes, experience with and perceptions of VHA care.

This paper reports the process by which the Veterans Engagement with Technology Collaborative cohort was developed, describes the baseline cross-sectional survey data collected from cohort members, and details future plans for longitudinal follow-up and cohort maintenance.

Methods

Design

The Veterans Engagement with Technology Collaborative cohort is a longitudinal cohort comprised of survey data collection across multiple time-points.

Participants

Individuals were eligible for inclusion in the cohort if they were a veteran of the US military and an active user of VHA health care services. In addition, because we aimed to enroll veterans who were users of patient-facing eHealth technology, at the time of screening, all veterans invited to be in the cohort were required to have a mobile phone and have sent at least 5 (but less than 30, which comprised the cut-off for the 95th percentile of secure message volume) secure messages using the patient-to–clinical team secure messaging feature of the VHA personal health record portal in the year prior to recruitment.

Setting

The VHA health care system provides health care and other benefits (such as compensation or pension, life insurance, and vocational rehabilitation) to approximately 9.7 million veterans of the US military, which is nearly half of the entire veteran population [15]. On average, compared to civilians, veterans are older, predominantly male, and experience more health concerns, and compared to veterans who do not receive VHA care, veterans who use VHA services tend to be sicker and have less income [16]. The veterans who currently comprise the Veterans Engagement with Technology Collaborative cohort received care from at least 1 of 14 purposefully sampled VHA medical centers across the United States and include residents of most states across both urban and rural settings. In addition to securing a diverse geographic representation of veterans, as well as adequate representation of women and racial or ethnic minority veterans, we selected these 14 facilities based on several criteria deemed important to the overall goals of the Veterans Engagement with Technology Collaborative cohort. Criteria included engaging veterans seen for care at VHA facilities representing different geographic regions of the country that had high rates of adoption of the VHA's personal health record portal secure messaging feature, a track record of being a site of early adoption for other VHA patient-facing eHealth technologies (eg, VHA's automated text messaging system, video-to-home telehealth, online scheduling), and plans for implementing other VHA eHealth technologies in the future. Veterans who met these criteria were included on recruitment lists, which detailed their name and contact information (eg, telephone number) as listed in VHA administrative records housed in the VHA Corporate Data Warehouse.

Recruitment

In order to recruit veterans into the cohort, we provided evaluation team members with these recruitment lists, and team members reached out to each veteran by phone to invite them to participate. We called all eligible patients one time unless they requested a call back, in which case, we made one additional follow-up call. During the call, team members read a script to the veterans explaining the purpose of the Veterans Engagement with Technology Collaborative cohort and their eligibility to participate. Evaluation team members also explained the long-term commitment requested of each participating veteran. That is, veterans who consented to participate in the cohort during this recruitment call would be engaged in ongoing survey efforts and periodic evaluation activities over time. If the veteran agreed to participate, the evaluation team member then collected the baseline survey data. Responses were entered in real time, into a secure electronic database system (REDCap, Research Electronic Data Capture; Vanderbilt University). Survey administration took approximately 20 minutes per patient. All recruitment calls and survey data collection efforts occurred in the 2017 calendar year. No incentives were provided at baseline data collection.

Baseline Data Collection

Survey Measures

The Veterans Engagement with Technology Collaborative baseline survey was used to gather patient-level information that is not readily available in clinical or administrative databases, such as patient perceptions of access to and current use of health care and patient-facing technologies, perceptions of health care team member support of these technologies, patient-provider communication, and sociodemographic information such as health literacy and financial status.

We asked participants to report on factors associated with their health and health care use, including whether they usually receive health care from the VHA, from outside of the VHA, or both, and how long it takes to travel from their home to their VHA primary care doctor's office. We also asked them whether anyone helps them manage their health or health care, how they perceive their overall health status [17], and the extent to which they adhere to taking their prescribed medications [18].

In addition, several questions addressed health-related goal setting behaviors; specifically, participants were asked, "In the last 6 months, did anyone in your VHA provider's office talk with you about specific goals for your health?" and "In the past 6 months, have you set any goals related to your health?" Those who set a goal were asked, "What health-related goal or goals have you made in the past 6 months?"; "Have you been able to achieve this health-related goal?"; and "Have you used an app on a smartphone or tablet to help you achieve/work on this health-related goal?"

Technology Ownership and Use

Technology ownership and use questions assessed personal computer ownership, tablet computer ownership, mobile phone ownership, whether participants ever borrow any of these devices from others, and whether participants ever use devices to measure and send health information (eg, blood pressure, blood glucose level, weight) to their care providers. We also asked participants whether they like to be among the first to get a new device, tech gadget, or app when it comes out (ie, do they consider themselves early adopters), whether they use social media (ie, Twitter, Facebook, Instagram, Pinterest) [19], and how comfortable or confident they feel using computers on a scale of 0 (not at all) to 5 (very) [20].

We used adapted items from the VHA Survey of Healthcare Experience of Patients to assess participant perceptions of their health and health care–related communication, including how easy it is for them to communicate with their care providers when needed on a scale of 1 (very easy) to 5 (very difficult), how often (in the prior 6 months) they received a response within 1 day when they needed to communicate with their care provider's office on a scale of 1 (never) to 4 (always), and how often (in the prior 6 months) they received a response from their care provider's office as soon as they needed it when they contacted the office after hours on a scale of 1 (never) to 4 (always).

We also asked participants how big of a problem on a scale of 1 (very big problem) to 5 (not a problem) each of the following are for them: poor communication between different doctors or clinics, disagreements between their doctors about their diagnosis or the best treatment for them, and having their concerns ignored or overlooked by their health care providers. In addition, we asked participants to report how confident they feel filling out medical forms by themselves on a scale of 1 (not at all) to 5 (extremely) [21], and how easy or hard they find it to understand medical statistics on a scale of 1 (very easy) to 4 (very hard) [22].

Additionally, we asked participants to report their agreement on a scale of 1 (strongly disagree) to 7 (strongly agree) with a number of statements about secure messaging: their health care team encourages them to ask questions using secure messaging; in secure messaging, their health care team answers their questions related to their health fully and carefully; and the secure messages they receive make them feel that their health care team cares about them as a person.

We collected demographic information including age, gender, race, ethnicity, relationship status, highest level of education achieved, living arrangement, and financial difficulty.

Data Linked From VHA Records

We obtained data on chronic health conditions and information used to calculate a Hierarchical Condition Community [23,24] score for the veterans in our cohort from the VHA Corporate Data Warehouse. Hierarchical Condition Community scores represent a comorbidity index that takes into account an individual's age, gender, medical diagnoses, and eligibility for Medicare and Medicaid services [23,24]. Typically, the range of Hierarchical Condition Community scores is between 0.9 and 1.7; scores less than 1 are often interpreted as healthy [25].

Statistical Analyses

We will use the data provided by the cohort to examine novel and important issues related to technology use among veterans, including perceptions of newly developed patient-facing technologies, impacts of use on perceptions of and satisfaction with care delivery, and associations with important health and utilization outcomes (eg, health-related goal setting and attainment, medication adherence, communication with care team members). In this manuscript, we examine frequencies of responses to key survey items. Statistical analyses were conducted using STATA (version 14.2; StataCorp LLC).

Results

Responses to survey items intended to characterize the sample and gather information on covariates are presented below in the narrative and accompanying tables.

Response Rate and Cohort Derivation

We identified and attempted to contact 20,091 veterans who met inclusion criteria for the cohort. Of these veterans, 5877 were reached by phone, 2735 agreed to participate, and 2727 completed the survey (46.4% participation rate).

Demographics

The veterans who comprised the cohort were 59.9 years old, on average, at the time that the first survey was administered in 2017. Participants were predominantly male (2268/2727, 83.2%), White (2226/2727, 81.6%), and living in their own apartment or house (2519/2696, 93.4%). Most had completed some college (1176/2701, 43.5%) or an advanced degree (1178/2701, 43.6%) and were married or in a civil union (1734/2687, 64.5%). Most (1813/2637, 68.8%) reported that it was not very difficult for them to pay for basics like food and heating or cooling (Table 2).

Table 2. Demographics.

Variable	Value
Age (years) (N=2727)	
mean (SD)	59.9 (13.1)
range	24.5-95.8
Gender (N=2727), n (%)	
Male	2268 (83.2)
Female	459 (16.8)
Race (N=2727), n (%)	
White	2226 (81.6)
Black or African American	317 (11.6)
Asian	13 (0.5)
Native Hawaiian or other Pacific Islander	6 (0.2)
American Indian or Alaskan Native	63 (2.3)
Other	86 (3.2)
Declined to answer	56 (2.1)
Ethnicity (N=2727), n (%)	
Yes, Hispanic or Latino	135 (5.0)
No, not Hispanic or Latino	2592 (95.1)
Relationship status (n=2687), n (%)	
Married or in a civil union	1734 (64.5)
Neither married, nor in a civil union[a]	953 (35.5)
Education status (n=2701), n (%)	
High school graduate or less	347 (12.9)
At least some college or vocational school (1-4 years)	1176 (43.5)
Master’s, professional, or doctoral degree	1178 (43.6)
Living arrangement (n=2696), n (%)	
Own apartment or house	2519 (93.4)
Friend or relative’s apartment or house	120 (4.5)
Other[b]	57 (2.1)
Financial difficulty[c] (n=2637), n (%)	
Not very hard	1813 (68.8)
Somewhat hard, hard, or very hard	824 (31.3)

[a] Defined as engaged or in a relationship, single, separated, divorced, or widowed.

[b] Defined as school or dormitory, hospital or detox center, nursing home or assisted living, car or street, or jail or prison.

[c] Based on response to the question “How hard is it for you (and your family) to pay for the very basics like food and heating/cooling?”

Health and Health Care Use

The most prevalent chronic conditions among this sample were hypertension (1699/2727, 62.3%), osteoarthritis (1444/2727, 53.0%), and depression (1109/2727, 40.7%). Most participants reported that they receive their health care mostly at the VHA (2142/2718, 78.8%), and nearly half (1326/2720, 48.8%) reported living less than 30 minutes away from the VHA at which they received primary care. The majority of the cohort reported being in good (1055/2725, 38.7%) or very good (524/2725, 19.2%) health and that they always take their medications as recommended by their care providers (2273/2698, 84.3%) (Table 3).

Table 3. Health and health care use.

Variable	Value
Chronic conditions[a] (N=2727), n (%)	
Acute myocardial infarction	297 (10.9)
Atrial fibrillation	227 (8.3)
Heart failure	206 (7.6)
Ischemic heart disease	646 (23.7)
Peripheral vascular disease	352 (12.9)
Hypertension	1699 (62.3)
Asthma	575 (21.1)
Breast cancer	23 (0.8)
Colorectal cancer	35 (1.3)
Prostate cancer	105 (3.9)
Lung cancer	32 (1.2)
Endometrial cancer	2 (0.1)
Chronic kidney disease	646 (23.7)
Chronic obstructive pulmonary disease	105 (3.9)
Depression	1109 (40.7)
Diabetes	1024 (37.6)
Osteoarthritis	1444 (53.0)
Stroke	160 (5.9)
Posttraumatic stress disorder	773 (28.4)
Anxiety	705 (25.9)
Traumatic brain injury	205 (7.5)
Health status (general) (n=2725), n (%)	
Excellent	144 (5.3)
Very good	524 (19.2)
Good	1055 (38.7)
Fair	797 (29.3)
Poor	205 (7.5)
Hierarchical Condition Community score[a] (N=2727), n (%)	
Mean (SD)	0.3 (0.03)
Range	0.3-0.4
Health care receipt (n=2718), n (%)	
Mostly at the VHA[b]	2142 (78.8)
Mostly outside VHA	159 (5.9)
About half in VHA, half outside VHA	417 (15.3)
Travel time (to VHA primary care doctor’s office) (n=2720), n (%)	
<30 minutes	1326 (48.8)
31 to 60 minutes	970 (35.7)
>60 minutes	424 (15.6)
Assistance in managing health or health care (N=2727), n (%)	
Paid caregiver	92 (3.4)

Variable	Value
Spouse/partner	694 (25.5)
Children	157 (5.8)
Family or extended family member	176 (6.5)
Friend	100 (3.7)
Other	29 (1.1)
N/A[c]	1717 (63.0)
Medication adherence[d] (n=2698), n (%)	
All (100%) of the time	2273 (84.3)
Not all of the time	425 (15.8)

[a] In the prior five years.

[b] VHA: Veterans Health Administration.

[c] N/A: not applicable.

[d] Based on response to the question “In the past month, how often did you take your medications as the doctor prescribed?”

Technology Ownership and Use

The majority of participants reported owning a personal computer (2609/2725, 95.7%), tablet computer (1616/2716, 59.5%), or smartphone (2438/2722, 89.6%). Most (2412/2727, 88.5%) reported that they do not borrow technological devices from others. Most reported that they agree (669/2715, 24.6%) or strongly agree (813/2715, 29.9%) that they are an early adopter of new technology and that they are very comfortable or confident using computers (1878/2705, 69.4%) (Table 4).

Table 4. Technology ownership and use.

Variable	Value
Owns a desktop or laptop computer (n=2725), n (%)	
Yes	2609 (95.7)
No	116 (4.3)
Owns a tablet computer (iPad, Kindle Fire, etc) (n=2716), n (%)	
Yes	1616 (59.5)
No	1100 (40.5)
Mobile phone ownership[a] (n=2722), n (%)	
Smartphone[b]	2438 (89.6)
Nonsmartphone mobile phone	261 (9.6)
No mobile phone	23 (0.8)
Borrow devices from others (N=2727), n (%)	
Sometimes use friend's device	48 (1.8)
Sometimes use family member's device	167 (6.1)
Use device at work	77 (2.8)
Sometimes use library/senior center/hospital/other location's device	42 (1.5)
No	2412 (88.5)
Use of devices to measure and send health measurements to health care team (n=2717), n (%)	
Yes	679 (25.0)
No	2038 (75.0)
Early tech adopter (n=2715), n (%)	
Strongly agree	813 (29.9)
Agree	669 (24.6)
Neutral	692 (25.5)
Disagree	323 (11.9)
Strongly disagree	218 (8.0)
Social media use (N=2727), n (%)	
Twitter	552 (20.2)
Facebook	1955 (71.7)
Instagram	466 (17.1)
Pinterest	479 (17.6)
Comfort or confidence using computers (n=2705), n (%)	
Very comfortable or confident	1878 (69.4)
Less than very comfortable or confident	827 (30.6)

[a] Based on response to the statement "If you have multiple cell phones, select the one you use most often."

[b] iPhone, Android, Blackberry, Windows phone, Symbian, or some other type of smartphone.

Discussion

Veterans Engagement With Technology Collaborative Cohort and the Learning Health Care System

Evidence regarding the use and effectiveness of patient-facing technologies is accumulating [26-31], but considerable gaps remain. Given the abundance of new patient-facing technologies that are being (and will continue to be) developed, health care systems will face an ongoing challenge to determine if and how best these technologies can be used to support patients and improve health care quality. The concept of the learning health care system holds that "learning while doing" should be the penultimate goal of health care organizations and emphasizes the importance of appropriate infrastructure, data resources, and partnerships between stakeholders [32,33]. The learning health care system is predicated on the active collaboration among all participants in a system, underscoring how critical it is to engage stakeholders—including patients—in evaluation and implementation efforts [33]. What we have described in this paper is one initiative that is helping to accentuate the voice of the veteran in ongoing efforts to realize the vision of the learning health care system within VHA.

The Veterans Engagement with Technology Collaborative cohort directly engages veterans to understand the potential benefits and possible unintended consequences related to the patient-facing technologies that the VHA is developing and implementing. This new initiative provides a means for veterans and program evaluators to test these technologies on a timeline that more closely reflects their rapid development and evolution. It also supports the rapid evaluation of unexpected but significant changes in the health care system that may influence the role of technology in care delivery. The coronavirus disease 2019 (COVID-19) pandemic and the surge in patient-facing technology use that has accompanied it, is a case in point. The cohort reflects a strong partnership between operational entities within a large health care system and established members of its research and evaluation community to develop new infrastructure to support the broad goal of implementing and measuring the impacts of patient-facing eHealth technologies in practice.

The application and advancement of health- and health care–related technology has the potential to help revolutionize care, improve patient outcomes and satisfaction, and reduce health care costs [34]. Notably, the veterans described in this manuscript have made a commitment to participate in longitudinal follow-up, which will consist of follow-up survey efforts by our team over several years, the content and timing of which will be driven by the VHA's evaluation needs. Some question items and scales included on the baseline survey will be repeated, thus providing longitudinal data. We expect that other question items and scales will be added based on emergent priorities. Through our follow-up data collection efforts, this cohort of veterans will also serve as a resource to evaluate future technologies, such as new mobile health apps developed for use within the VHA to optimize important outcomes (eg, access to and coordination of services, patient activation and self-management, goal setting and attainment). Cohort member involvement in evaluations is likely to include providing feedback via multiple approaches including targeted surveys and interviews focused on user experiences. Evaluation activities that incorporate the voice of the veteran are increasingly recognized by the VHA health care system and its leadership as powerful approaches to improving health care delivery in ways that reflect the needs and preferences of the veteran population. The Veterans Engagement with Technology Collaborative cohort aligns with other veteran engagement initiatives currently being implemented to improve the policy and patient relevance of VHA research and evaluation activities.

Limitations

Survey responses are subject to a number of biases (eg, recall bias, response bias), and the cross-sectional design does not allow us to determine causal relationships. Furthermore, the veterans in our sample represent a subset of the veteran population and are known technology users, and in comparison to the general population of veterans who use VHA health care, are approximately one year younger, on average [35], and include a greater proportion of women [36], individuals who are White and of non-Hispanic ethnicity, individuals who report being in fair or poor health [35], and individuals who have health conditions such as depression [37,38], diabetes [39], posttraumatic stress disorder [38,40], hypertension [41], and anxiety [38]. These differences may impact patient-facing eHealth technology preferences or use and limit the generalizability of the data collected thus far, as well as findings from future evaluations conducted with this cohort.

We also acknowledge that the proxy indicators used for technology adoption (ie, veteran use of secure messaging in the prior year, early implementation of eHealth technologies at VHA facilities where veterans receive health care) do not comprehensively reflect all factors that may impact use of patient-facing eHealth technologies. Future work may consider additional factors, for example, aspects of the technology's design, usability, and utility. In addition, while we recognize the importance of health care providers and their perspectives in the development and evaluation of patient-facing technologies, we have not yet incorporated their perspectives into the initiative. Because all patient-facing technologies have reciprocal repercussions for the health care team members of patients who use them, in the future, we also plan to assess the perspectives of VHA health care team members.

Conclusions

Through the development of the Veterans Engagement with Technology Collaborative cohort, the VHA is laying the foundation for future, rapid, rigorous evaluation and quality improvement efforts that can advance our understanding of the adoption, use, and impact of patient-facing technologies and inform related policy decisions and funding priorities. The development and maintenance of the Veterans Engagement with Technology Collaborative cohort over time establishes a diverse group of veterans who can test emerging VHA patient-facing technologies and technology-based interventions. This infrastructure will help us obtain early feedback on these technologies, as well as advance our understanding of whether certain groups of veterans require extra support to adopt these technologies and use them over time.

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Conflicts of Interest

None declared.

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Abbreviations

mHealth: mobile health
VHA: Veterans Health Administration

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Original Paper

Perceived Need for Psychosocial Support After Aortic Dissection: Cross-Sectional Survey

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Abstract

Background: The gold standard management of aortic dissection, a life-threatening condition, includes multidisciplinary approaches. Although mental distress following aortic dissection is common, evidence-based psychosocial interventions for aortic dissection survivors are lacking.

Objective: The aim of this study is to identify the perceived psychosocial needs of aortic dissection survivors by surveying patients, their relatives, and health professionals to inform the development of such interventions.

Methods: This study used a cross-sectional survey and collected responses from 41 participants (27 patients with aortic dissection, 8 relatives of patients with aortic dissection, and 6 health professionals) on key topics, types of interventions, best timing, anticipated success, and the intended effects and side effects of psychosocial interventions after aortic dissection.

Results: The principal intervention topics were “changes in everyday life” (28/41, 68%, 95% CI 54.5%-82.9%), “anxiety” (25/41, 61%, 95% CI 46.2%-76.2%), “uncertainty” (24/41, 59%, 95% CI 42.9%-73.2%), “tension/distress” (24/41, 59%, 95% CI 43.9%-73.8%), and “trust in the body” (21/41, 51%, 95% CI 35.9%-67.5%). The most commonly indicated intervention types were “family/relative therapy” (21/41, 51%, 95% CI 35%-65.9%) and “anxiety treatment” (21/41, 51%, 95% CI 35%-67.5%). The most recommended intervention timing was “during inpatient rehabilitation” (26/41, 63%, 95% CI 47.6%-77.5%) followed by “shortly after inpatient rehabilitation” (20/41, 49%, 95% CI 32.4%-65%). More than 95% (39/41) of respondents anticipated a benefit from psychosocial interventions following aortic dissection dissection, expecting a probable improvement in 68.6% (95% CI 61.4%-76.2%) of aortic dissection survivors, a worse outcome for 5% (95% CI 2.9%-7.9%), and that 6% (95% CI 1.8%-10.4%) would have negative side effects due to such interventions.

Conclusions: Our findings highlight a substantial need for psychosocial interventions in aortic dissection survivors and indicate that such interventions would be a success. They provide a basis for the development and evaluation of interventions as part of state-of-the-art aortic dissection management.

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KEYWORDS

aortic dissection; patient involvement; psychosocial support; psychosomatic; psychotherapy; treatment need

Introduction

Aortic dissection is a rare but life-threatening condition. The annual incidence of the condition in older adults is up to 35 cases per 100,000 people, with a slight male preponderance [1-3]. This low incidence explains the delayed diagnosis; only 39% of patients are diagnosed within 24 hours after symptom onset [4]. Prognosis is grave, with a lethality rate of 1% to 2% per hour after onset of symptoms in untreated patients [5]. Preadmission mortality is 20% [6]. Operative 30-day mortality for ascending aortic dissection at experienced centers is still between 10% and 35% [4]. In a propensity-matched retrospective analysis, survival rates in patients with acute type A dissection were 91% after 30 days, 74% after 1 year, and 63% after 5 years [4]. Furthermore, 10-year survival rates of patients who are discharged from hospital range from 30% to 60% [6]. The underlying pathophysiology of aortic medial disease and defective wall structure confers an ongoing risk of further dissection, aneurysmal degeneration, and rupture. Therefore, consequent control of known risk factors is crucial. In addition to age and genetic disorders such as Marfan syndrome, risk factors include lifestyle factors such as long-term arterial hypertension, smoking, dyslipidemia, and cocaine, crack cocaine or amphetamine substance use [3]. All this can be highly traumatic and stressful for affected patients, their relatives, and health care professionals.

Contemporary management of aortic dissection should be multidisciplinary and include, among other things, serial noninvasive imaging, biomarker testing, genetic risk profiling for aortopathy, blood pressure and heart rate control, lipid-lowering therapies, and repairing or replacing the damaged region of the aorta [2,3,7], with evidence that successful surgical intervention substantially improves the quality of life of aortic dissection survivors [8-10].

With regard to psychosocial factors, previous recommendations focus on patient education and the achievement of lifestyle goals. They include ensuring adherence to medical treatment, genetic counselling, smoking cessation, and other risk factor modification for atherosclerotic disease, as well as avoidance of cocaine or other stimulating drugs [2]. Furthermore, counselling aortic dissection survivors on exercise and physical activity has been described as important yet challenging, given the need to ensure avoidance of potentially damaging strenuous physical activities and contact sports, while fostering moderate intensity cardiovascular activity that may be cardioprotective in this patient cohort [11].

Initial evidence showed changes in lifestyle and emotional state following aortic dissection, with physical and sexual activity decreasing slightly, the latter mostly in relation to fear, while approximately one-third of aortic dissection survivors reported new subjective feelings of depression and anxiety [12]. However, anecdotal reports suggest that regular provision of psychosocial support to aortic dissection survivors, including psychotherapy and other interventions for psychosocial distress or mental disorders, is scarce.

Despite common clinical observations of psychosocial distress related to aortic dissection, to the best of our knowledge, there is no evidence-based tailored psychosocial (ie, psychosomatic or psychotherapeutic) intervention for aortic dissection survivors, or even systematic information regarding the need for psychosocial interventions following aortic dissection. Therefore, as a first step toward the development of a tailored psychosocial support intervention for aortic dissection survivors, we conducted a survey, addressing aortic dissection survivors, relatives of aortic dissection survivors, and professionals working with patients with aortic dissection.

The main aims of this survey were to systematically identify (1) the key topics that need psychosocial attention for aortic dissection survivors, (2) the main types of desired psychosocial interventions, (3) the expected success and both the intended and unintended effects of such interventions, and (4) the preferred timing of such interventions.

Our study is in line with recent efforts to involve patients early in the development of new interventions, which has become a key issue in biomedical research (eg, see the British Medical Journal’s Partnering with Patients initiative (“nothing about us without us”) [13]. According to this movement, partnering with patients, their families, support communities, and the public is an ethical imperative, which is essential to improving the quality, safety, value, and sustainability of health care systems.

Methods

Study Setting and Design

We present the results from a cross-sectional survey administered during an aortic dissection information event (Aortic Dissection Awareness Day 2017) to aortic dissection survivors and their families and relatives, as well as health professionals dealing with this condition at the University Hospital Basel.

Ethical clearance for this study was acquired from the Ethikkommission Nordwest- und Zentralschweiz (EKNZ) in Basel, Switzerland (EKNZ BASEC Req-2017-00916). Each participant provided consent that his/her responses could be used for analyses that would be reported in scientific publications.

We describe the results of our survey based on the Guidance for Reporting Involvement of Patients and the Public (GRIPP) 2, the first international guidance for reporting of patient and public involvement in health and social care research [14]. The GRIPP2 short form checklist short form accompanying this article is provided as Multimedia Appendix 1.

Study Instrument

The questionnaire we used was a self-developed instrument compiled by an interdisciplinary team. The contents of the questionnaire, including the response categories, were compiled over the course of several meetings on the basis of pertinent publications [12,15] and information obtained during clinical encounters with patients with aortic dissection. We added open response options to ensure that respondents could provide replies beyond the preselected categories. Draft versions of the questionnaire were circulated and, based on written feedback and exchanges during another meeting, the instrument was modified until consensus was reached. This resulting questionnaire covers respondents' sociodemographic information, topics that may require psychosocial attention for aortic dissection survivors and their relatives, types of psychosocial interventions preferred, the expected intended and unintended effects and anticipated success of such interventions, and the preferred timing of such interventions. Furthermore, we asked the respondents to indicate the percentage of aortic dissection survivors they knew that had received psychosocial support or interventions, and to specify the support or interventions. Finally, we left space for additional comments.

Answer formats were predominantly prespecified response categories that allowed for additional open responses. Other questions used a 7-point Likert-scale ("strongly disagree" to "strongly agree"), or asked for frequencies expressed as percentages.

A professional translator ensured equivalence of the French and German versions of the questionnaire. An English translation of the questionnaire is provided as Multimedia Appendix 2. The German and French versions are available from the authors on request.

Recruitment of Respondents

During an aortic dissection information event, Aortic Dissection Awareness Day 2017, we administered a paper-pencil version of our questionnaire in German and French, enabling anonymous responses. The event was open to the public and participants included aortic dissection survivors and their families and relatives, as well as health professionals dealing with this condition. All attendees were invited to participate in the survey, without any predetermined number of participants being asked to complete the survey. We included responses from aortic dissection survivors, their families and relatives, and health professionals.

Analyses and Statistics

We analyzed the responses by using descriptive statistics, calculating means and frequencies, and estimating 95% confidence intervals (95% CIs), based on bootstrapping procedures (1000 repetitions), to provide a measure of accuracy to our findings in terms of estimates. We did not apply the bootstrapping procedure in cases where the subgroup sample size was insufficient to calculate meaningful CIs (in these cases, no 95% CIs are provided). We provide analyses for the total sample and stratified analyses according to (1) aortic dissection survivors, (2) relatives of aortic dissection survivors, and (3) health professionals concerned with patients with aortic dissection. Open answers and comments were evaluated according to the principles of Qualitative Content Analysis [16]. Statistical analyses were performed using IBM SPSS Statistics for Macintosh (Version 21.0, IBM Corp).

Results

Study Sample Characteristics

A total of 41 aortic dissection survivors, their families and relatives, and health professionals participated in the study and consented to the use of their information for research purposes. In addition, 3 participants (2 partially, 1 fully) completed the questionnaire without consenting to the use of their information for research purposes. Data from these 3 subjects (7% of eligible respondents) and data from 3 noneligible respondents (who did not fall under one of the 3 subgroups surveyed in this study; 6% of all respondents) were excluded from analyses. The majority of the 41 subjects included in the study were patients with aortic dissection (n=27), followed by relatives of patients with aortic dissection (n=8), and health professionals (n=6). The characteristics of the study sample are presented in the table below (Table 1).

Table 1. Characteristics of the study sample.

Characteristics	Patients with aortic dissection (n=27), n (%)	Relatives of patients with aortic dissection (n=8), n (%)	Health professionals (n=6), n (%)	All subjects (n=41), n (%)
Gender				
Male	21 (84)	0 (0)	4 (67)	25 (69)
Female	4 (16)	5 (100)	2 (33)	11 (31)
No response[a]	2	3	N/A[b]	5
Age (years)				
<30	1 (4)	0 (0)	2 (33)	3 (7)
30-39	0 (0)	1 (13)	0 (0)	1 (2)
40-49	3 (11)	0 (0)	3 (50)	6 (15)
50-59	7 (26)	2 (25)	1 (17)	10 (24)
60-69	10 (37)	4 (50)	0 (0)	14 (34)
>69	6 (22)	1 (13)	0 (0)	7 (17)
Language				
German	26 (96)	8 (100)	6 (100)	40 (98)
French	1 (4)	0 (0)	0 (0)	1 (2)
Number of other patients with aortic dissection in contact with in a year				
None	17 (68)	2 (25)	1 (17)	20 (51)
1-3 patients	8 (32)	6 (75)	2 (33)	16 (41)
4-8 patients	0 (0)	0 (0)	0 (0)	0 (0)
9-19 patients	0 (0)	0 (0)	0 (0)	0 (0)
20-40 patients	0 (0)	0 (0)	1 (17)	1 (3)
≥40 patients	0 (0)	0 (0)	2 (33)	2 (5)
No response[a]	2	N/A	N/A	2

[a] Where participants did not respond to a question, they were not counted in the percentage calculations.

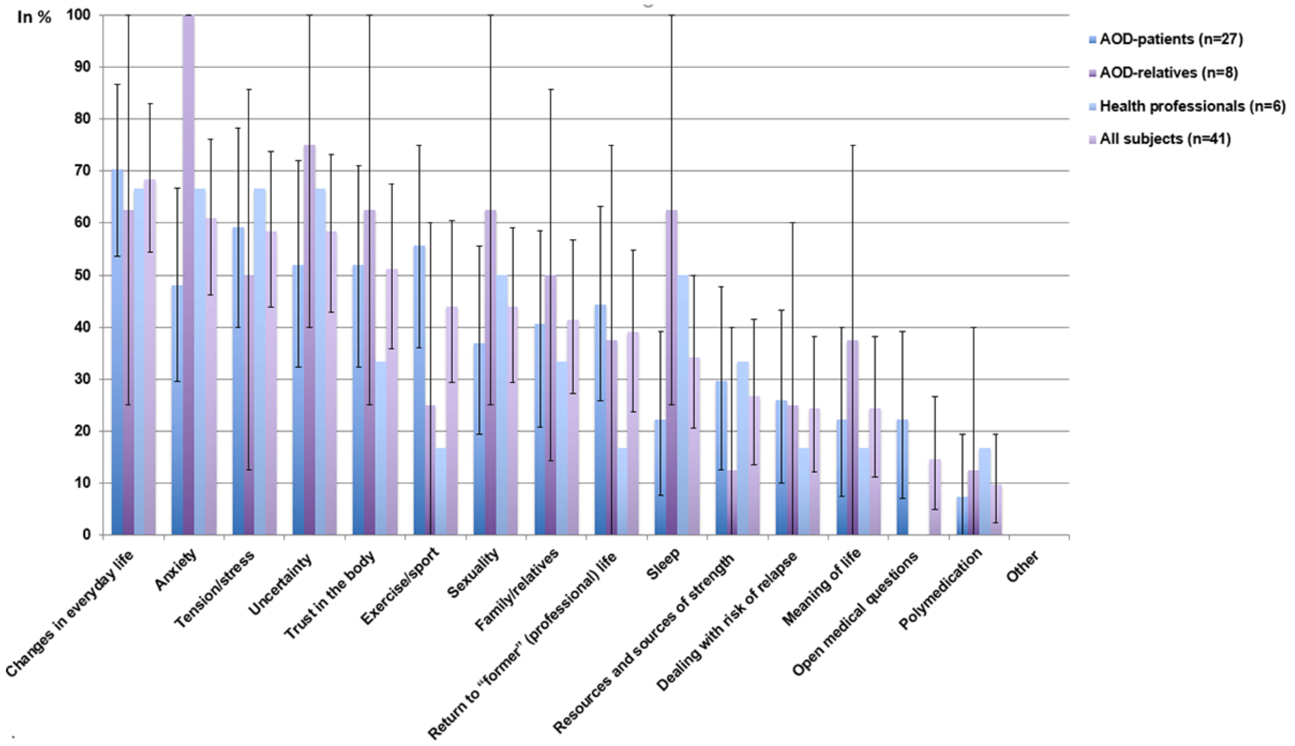
[b] N/A: not applicable.

Key Topics

On average, 6 topics (95% CI 5.2-6.9; Table 2) that need psychosocial attention after aortic dissection were indicated per respondent. The most common topics (Figure 1), all chosen by more than half of the respondents, were “changes in everyday life” (28/41, 68%, 95% CI 54.5%-82.9%), “anxiety” (25/41, 61%, 95% CI 46.2%-76.2%), “uncertainty” (24/41, 59%, 95% CI 42.9%-73.2%), “tension/stress” (24/41, 59%, 95% CI 43.9%-73.8%), and “trust in the body” (21/41, 51%, 95% CI 35.9%-67.5%). The only additional topics in the free response category were “insurance issues” and “pension issues” mentioned by 1 aortic dissection survivor.

Compared to aortic dissection survivors, relatives of patients with aortic dissection indicated “anxiety,” “sexuality,” and “sleep” more often and “open medical questions” and “exercise/sport” less often (none indicated “open medical questions”). Health care professionals indicated “exercise/sport,” “return to former (professional) life,” and “open medical questions” less often than aortic dissection survivors (none indicated “open medical questions”).

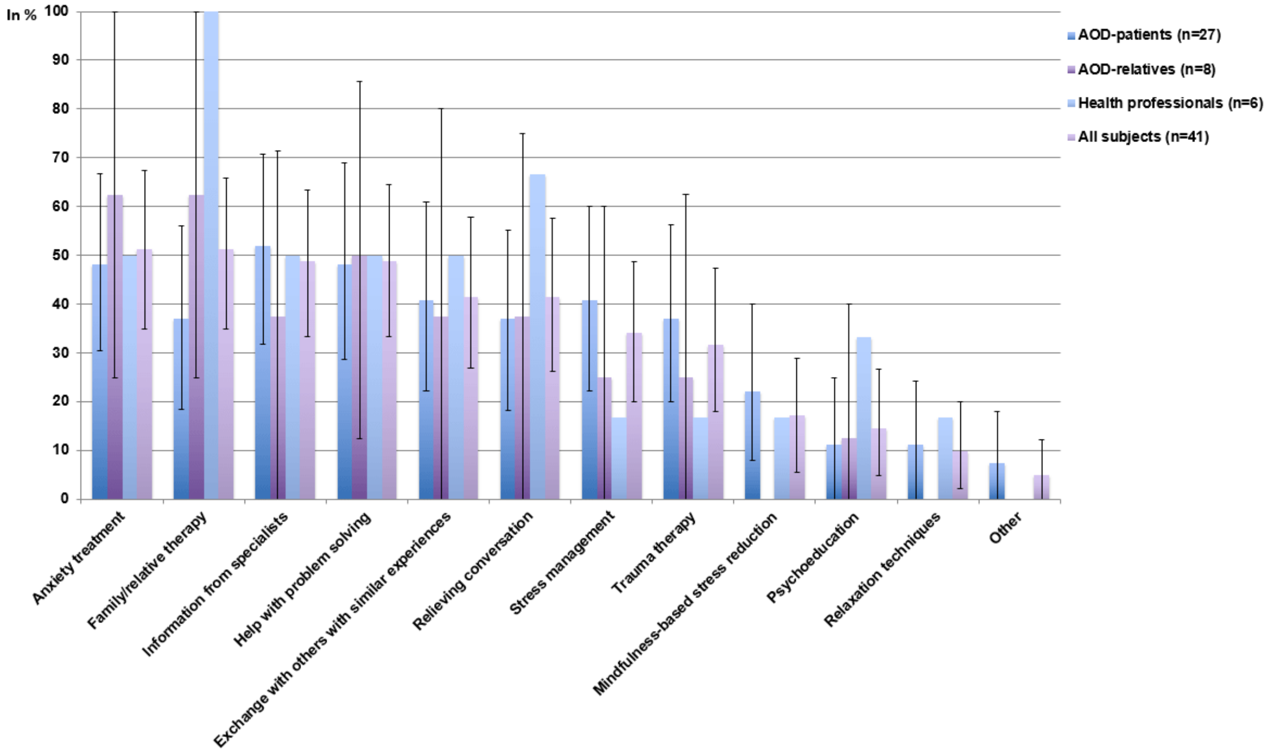
Figure 1. Frequency of subjects mentioning a certain concern or topic of interest to aortic dissection survivors to be addressed by psychosocial-psychotherapeutic interventions (mean and 95% CI). AOD: aortic dissection.



Types of Psychosocial Interventions Preferred

On average, each respondent indicated 4 different types (95% CI 3.3-4.7) of desired psychosocial interventions for aortic dissection survivors. The most common types (Figure 2), indicated by more than half of the respondents, were "family/relative therapy" (21/41, 51%, 95% CI 35%-65.9%) and "anxiety treatment" (21/41, 51%, 95% CI 35%-67.5%). In addition, 2 aortic dissection survivors used the free response option to add "help with clarifications regarding insurance and pension issues" and "obtaining a better understanding of what residual impairment is 'normal' after aortic dissection."

Figure 2. Frequency of subjects mentioning a certain type of psychosocial-psychotherapeutic intervention being of relevance to aortic dissection survivors (mean and 95% CI). AOD: aortic dissection.

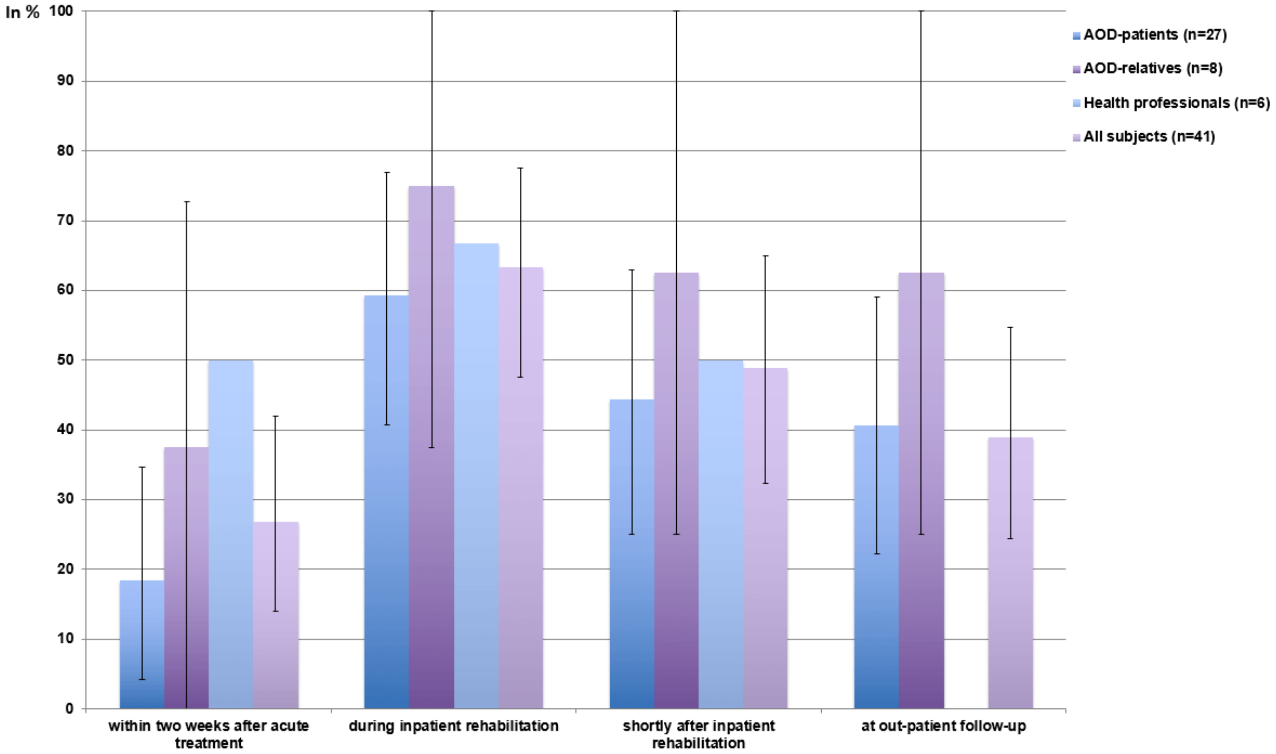


Compared to aortic dissection survivors, relatives indicated “family/relative therapy” more often and “mindfulness-based stress reduction” (MBSR) and “relaxation techniques” less often (not at all). Compared to aortic dissection survivors, health care professionals chose “family/relative therapy,” “relieving conversation,” and “psychoeducation” more often, and “stress-management” and “trauma therapy” less often.

Best Timing of Intervention

The most commonly chosen appropriate time to propose psychosocial support after aortic dissection was “during inpatient rehabilitation” (26/41, 63%, 95% CI 47.6%-77.5%) followed by “shortly after inpatient rehabilitation” (20/41, 49%, 95% CI 32.4%-65%), “at outpatient follow-up” (16/41, 39%, 95% CI 24.4%-54.7%), and “within two weeks after acute treatment” (11/41, 27%, 95% CI 14%-41.9%; Figure 3).

Figure 3. Frequency of subjects proposing a certain time as appropriate for psychosomatic or psychotherapeutic support after aortic dissection (mean and 95% CI). AOD: aortic dissection.



Anticipated Success

More than 95% (39/41) of respondents “strongly agreed” (20/41, 49%, 95% CI 33.3-64.3), “agreed” (12/41, 29%, 95% CI 15.9-44.5), or “somewhat agreed” (7/41, 17%, 95% CI 5.4-28.9) that patients would benefit from psychosocial support after aortic dissection, with relatives and health care professionals being slightly more optimistic than aortic dissection survivors (Figure 4; mean scores provided in Table 2).

Intended Effects and Side Effects

In total, a mean of 68.6% of respondents expected that aortic dissection survivors (95% CI 61.4-76.2) would improve due to psychosocial support, while a mean of 5.2% (95% CI 2.9-7.9) expected they would be worse off. Furthermore, a mean of 5.7% of aortic dissection survivors (95% CI 1.8-10.4) expected to experience negative side effects from psychosocial support (Table 2).

Types of expected side effects were only indicated by aortic dissection survivors and included the following: “other problems predominately,” “depression,” “stress,” “the fears are greater than the support suggests,” “egocentric manifestations,” “fear of relapse,” and “to focus too much on the disease when the ability to work is restored.”

Qualitative Findings

Answers to the open question “What else would you consider important to improve psychosomatic or post–aortic dissection psychotherapeutic support?” were sorted according to the categories identified and are provided in Table 3. The majority of responses can be subsumed under the categories “emotional support and encouragement” and “information, counsel, and assistance.”

Figure 4. Frequency of subjects agreeing that patients would benefit from psychotherapeutic support after aortic dissection (mean and 95% CI). AOD: aortic dissection.

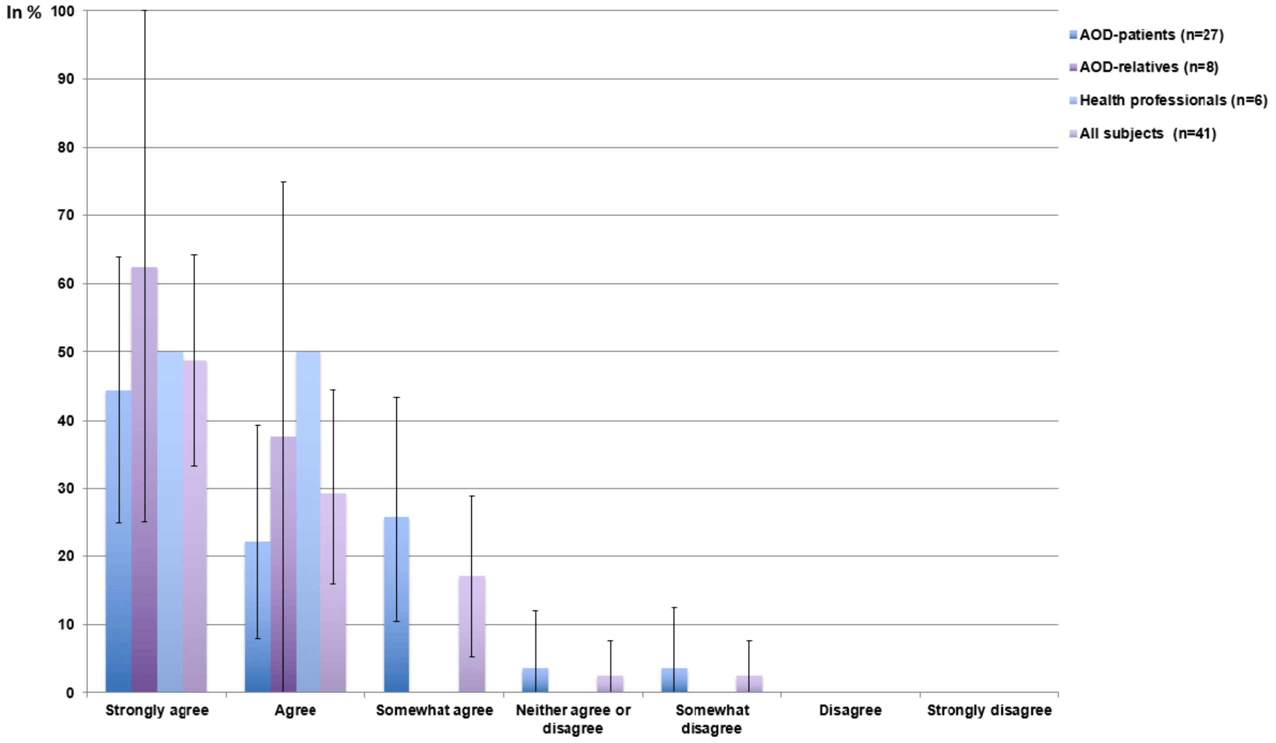


Table 2. Survey results and continuous variables.

Variable	Scale	Patients with aortic dissection (n=27), mean (95% CI)	Relatives of patients with aortic dissection (n=8), mean (95% CI)[a]	Health professionals (n=6), mean (95% CI)[a]	All subjects (n=41), mean (95% CI)
Extent of agreement with “Patient would benefit from interventions”	1 (strongly disagree) to 7 (strongly agree)	6 (5.57-6.4)	6.63 (6.25-7)	6.5 (6-7)	6.2 (5.9-6.5)
Expected frequency of aortic dissection survivors that improve	Percentages	66.7 (56.9-76.4)	70 (53.8-85.5)	75	68.6 (61.4-76.2)
Expected frequency of aortic dissection survivors that decline	Percentages	5.5 (2.9-8.2)	5 (5-5)	4.2	5.2 (2.9-7.9)
Expected frequency of aortic dissection survivors with negative side effects	Percentages	9.1 (3.4-16.4)	0	0.8	5.7 (1.9-10.4)
Frequency of acquaintances receiving psychosocial treatment	Percentages	2.2 (0-6.7)	16.7 (0-50)	35	12.5 (0.8-28.9)
Number of mentioned topics of aortic dissection survivors that need psychosocial attention per respondent	Absolute frequency	5.9 (4.7-7.1)	6.8 (5.6-8)	5.5	6 (5.2-6.9)
Number of mentioned types of desired psychosocial interventions per respondent	Absolute frequency	3.9 (3.1-4.8)	3.5 (2.5-4.5)	4.7	4 (3.3-4.7)

[a] 95% CI was calculated given sufficient sample size and distribution of values.

Table 3. Open answers to the question "What else would you consider important to improve psychosomatic or post–aortic dissection psychotherapeutic support?"

Type of respondent and response	Category
Patients with aortic dissection	
Encouraging conversation	Emotional support, encouragement
Being accompanied	Emotional support, encouragement
That one deals with the patient intensively and really does not give up	Emotional support, encouragement
I was supported by [*name of physician*]. That carried me through the operation and recovery	Emotional support, encouragement
To help people who need invalidity insurance after aortic dissection, in the fight with the insurance company and the pension fund	Information, counsel, assistance
Support with invalidity insurance	Emotional support, encouragement
Family support	Family
Inclusion of relatives and starting during rehabilitation	Family
That the option is offered and that relatives can access help both now and later	Family
With mechanical heart valve (how to deal with the sound)	Information, counsel, assistance
That a consultation is offered as early as possible. Similar to care teams after suicide or comparable events. I had to fight to receive a psychological conversation during rehabilitation. Thereafter, psychotherapy.	Information, counsel, assistance
Assistance in dealing with AHV/IV (pension insurance invalidity insurance)/pension fund, etc. Clarification of financial situation.	Information, counsel, assistance
Workplace: What work can I still do?	Information, counsel, assistance
To provide necessary information (addresses) of contacts after leaving the hospital (eg, cardiologist, psychologist, etc)	Information, counsel, assistance
To inform patients and their relatives early regarding opportunities for support	Information, counsel, assistance
You should go back to "everyday life" but still take care	Topics
Not getting good sleep	Topics
Reintegration into the work process is a challenge for indefinable reasons (difficult-to-explain symptoms)	Topics
Relatives of patients with aortic dissection	
Someone who listens to you and appreciates you!	Emotional support, encouragement
…that the relatives are involved. Too bad that this offer did not exist 5 years ago.	Family
Patient and relatives	Family
Show ways for the future; to reduce fear; to process the event; support with invalidity insurance	Emotional support, encouragement Information, counsel, assistance
To reduce fear by education	Information, counsel, assistance
Health professionals	
To reduce fear through better information	Information, counsel, assistance

Already Received Psychosocial Support

The average percentage of aortic dissection survivors that have received psychosocial support or treatment known to the respondents was 12.5% (95% CI 0.8%-28.9%; Table 2).

The type of support received included "relaxation exercises," "exercise/sport," "nutrition," "support from psychologist already received because of severe arthritis and claustrophobia," "self-organized after rehabilitation," "the family doctor provided a lot of support with conversation, etc," and "my partner was looking for support himself."

Discussion

In line with current patient involvement standards and to inform the development of psychosocial interventions, this study aimed to identify the needs of aortic dissection survivors, as indicated by aortic dissection survivors, their relatives, and health care professionals.

The most common topics to be addressed with such interventions included "disturbances in everyday life," "anxiety," "uncertainty," "tension/stress," and "trust in the body." The preferred types of interventions included "family/relative therapy" and "anxiety treatment." The top recommended intervention timing was "during inpatient rehabilitation" followed by "shortly after inpatient rehabilitation." Respondents anticipated that aortic dissection survivors would largely benefit from psychosocial interventions, on average expecting that approximately two-thirds would improve while only few would worsen or experience negative side effects.

Our study confirms the clinical impression that as current practice, only a minority of aortic dissection survivors receive psychosocial support, and if they do, it is limited to very few topics, such as exercise, sports, or anxiety.

Our findings are in line with and extend previous evidence on aortic dissection survivors, underlining the relevance of anxiety and uncertainty, topics related to activity, sports, and exercise, as well as sexuality after aortic dissection [12]. However, our findings also highlight hitherto unidentified topics, such as "trust in the body," "tension or stress," "everyday and professional life," as well as "family and relatives." Furthermore, the topics identified here are compatible with and expand reports on patients with unspecified life-threatening diseases in the context of intensive care units, with previous reports highlighting anxiety, uncertainty, and stress [17].

As the study comprised far fewer relatives and health professionals than aortic dissection survivors, conclusions based on comparisons between these groups' responses need to be drawn with caution. Nevertheless, our findings suggest that while there appeared to be a large overlap of views across these groups, there were also relevant differences (eg, compared to aortic dissection survivors, relatives mentioned anxiety and uncertainty more often as topics to address, and family/relative therapy as a desired intervention).

Strengths and Limitations

Our study has several strengths. We not only approached aortic dissection survivors and professionals, but also relatives of aortic dissection survivors, adding a perspective relevant to aortic dissection survivors' needs and expectations regarding psychosocial interventions. Furthermore, we assessed a broad spectrum of potential intervention topics and types, complemented by open questions, thereby embracing a broad range of needs regarding psychosocial interventions after aortic dissection. Our study also has several limitations: (1) By recruiting subjects as a convenience sample on an aortic dissection information day, we cannot exclude selection bias, potentially limiting the generalizability of our findings. However, the age and gender ratio of aortic dissection survivors participating in the survey was largely comparable to previous reports [18]. (2) The sample consisted primarily of aortic dissection survivors, and included a rather small number of relatives and health care professionals. All relatives that participated in the survey and provided gender information were female, highlighting the need to approach male relatives of aortic dissection survivors in future studies. (3) Given the moderate sample size, we did not stratify analyses with regard to age group, gender, or subtypes of aortic dissection. However, there is no clear rationale as to why a certain subtype of aortic dissection should have a risk profile different than that of other subtypes.

Implications for Research and Clinical Practice

Future studies should increase the number of respondents, including relatives of patients with aortic dissection as well as health professionals with different specializations (cardiologists, psychiatrists, etc); address additional questions, such as the preferred setting of psychosocial interventions (individual face-to-face versus group face-to-face versus online interventions); and expand the response categories to include additional topics, such as substance use.

The results of our study have important clinical implications. They guide the development of psychosocial interventions for aortic dissection survivors and their relatives in several ways: (1) the results highlighted the large number and substantial variety of issues that should be targeted, suggesting that a modular approach that allows a tailored and personalized compilation of intervention modules and techniques may be required to appropriately address diverse and complex individual needs; (2) they underscored the importance of potentially involving family and relatives and addressing related topics; (3) the results indicated potential undesired effects, such as iatrogenic fixation onto fears and the disease, hindering the focus necessary for restoration of participation in daily life and working ability; and (4) they suggested that interventions should be available along the whole disease period, from acute treatment directly after aortic dissection, to inpatient rehabilitation and the time thereafter, including outpatient follow-up.

We are currently establishing consultation-liaison psychosomatic support for patients with aortic dissection, informed by the findings reported here. To this end, we consider the individual needs of each patient and her or his family to develop interventions that fit each support-seeker best.

Conclusions

In this study, we described the results from a survey answered by aortic dissection survivors, their relatives, and health care professionals. These results provide a basis to inform the development of tailored psychosocial interventions. Overall, patient involvement was very well perceived and feasible, suggesting that it should become common practice when developing new psychosocial interventions in cardiology and beyond. Our findings highlight a substantial need and the anticipated success of psychosocial interventions for aortic dissection survivors and their relatives, and provide the basis for the development and evaluation of therapies that could become part of state-of-the-art aortic dissection management.

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Authors' Contributions

GM, RM, AF, SH, and RS conceptualized and designed the survey. GM and SG conducted statistical analyses. GM, RM, AF, SH, and RS interpreted data. GM wrote the initial manuscript. DB, RM, AF, SG, US, FSE, SH, and RS critically reviewed the manuscript. GM, DB, RM, AF, SG, US, FSE, SH, and RS approved the final version of the manuscript.

Conflicts of Interest

None declared.

Multimedia Appendix 1
GRIPP2 checklist.
[DOCX File , 16 KB - jopm_v12i3e15447_app1.docx]

Multimedia Appendix 2
Questionnaire.
[DOCX File , 642 KB - jopm_v12i3e15447_app2.docx]

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Abbreviations

EKNZ: Ethikkommission Nordwest- und Zentralschweiz
GRIPP: Guidance for Reporting Involvement of Patients and the Public
IPU: International Psychoanalytic University
SNSF: Swiss National Science Foundation

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