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Viewpoint

Bugs in the Virtual Clinic: Confronting Telemedicine's Challenges Through Empathy and Support

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Abstract

Although telemedicine has been an important conduit for clinical care during the COVID-19 pandemic, not all patients have been able to meaningfully participate in this mode of health care provision. Challenges with accessing telemedicine using consumer technology can interfere with the ability of patients and clinicians to meaningfully connect and lead to significant investments in time by clinicians and their staff. In this narrative case, we identify issues related to patients' use of technology, make comparisons between telehealth adoption and the deployment of electronic health records, and propose that building intuitive and supported digital care experiences for patients is required to make virtual care sustainable.

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Introduction

Background

"It's okay... just tell me what you see on your screen." A deep sigh punctuated with frustration was clearly audible across the phone line. It was a little after 6 PM. With the clinic staff having left an hour ago to begin their weekends, Ms J continued to struggle with her doctor to get connected to our telemedicine system for a virtual examination.

As informaticists who lead our health system's digital health program, we work to scale up virtual care capabilities and make them quickly accessible for patients and clinicians, who are all rapidly adapting to new challenges. Through experiences described here, we reflect on patient adoption of technology as a medium for care, draw similarities between the history of electronic health record (EHR) adoption and virtual care adoption, and contemplate how building meaningful digital care experiences for patients rather than creating digital carbon copies of traditional care is the way forward.

Ms J, a gregarious woman in her 70s with mild chronic obstructive pulmonary disease and well-controlled hypertension, had been scheduled for a telephone check-in earlier in the day. It was during the initial COVID-19 wave, when personal protective equipment was scarce and hospitals, including ours, were managing most patients virtually for the first time. The front desk staff had left a comment in the schedule: "Patient cannot do a video visit." As clinical informaticians, we had been struck by how fast health care transitioned to using video as the primary means for ambulatory care during the COVID-19 pandemic. And although our practice had been working to increase the number of video visits, clinicians defaulted to phone calls to provide care for those unable to access video visits, such as Ms J. As it turned out, Ms J's complaint over the phone was a new one, painful bumps on her skin "like bug bites but more purple," a problem for which at least a virtual exam would be warranted to begin the diagnostic process. Compared to a phone call, a video visit can allow for important nonverbal communication, further aiding the diagnostic and therapeutic process, such as the ability to confirm that both the patient and the clinician understand the information being shared through

facial expressions [1,2]. Without video, the differential diagnosis was too broad, ranging from bug bites to palpable purpura.

In the COVID-19 era, the need for physical distancing, the shortages of personal protective equipment, and the patchwork of state-issued guidance around stay-at-home orders had driven patients and providers alike to flock to virtual care. Insurance companies and the Centers for Medicare and Medicaid Services had also agreed to cover many telemedicine services, a step the federal government has committed to maintaining, including flexibility for audio-only visits for Medicaid beneficiaries [3,4]. With clear incentives for its use and the regulatory and reimbursement limitations historic to telemedicine dissolved at once, virtual care had now spread broadly across the country [5]. This represented the first real growth of telemedicine using consumer technology. Within our system, ambulatory care went from 0.25% to 70% virtual within 2 weeks of the pandemic. But as we see virtual care receding, locally accounting for 10% of all ambulatory care, now is the time to focus on creating sustainable virtual experiences following this large-scale “experiment.” We foresee a hybrid care model being adopted moving forward, meaning that clinical offices will intermix in-person and virtual care within the same clinical sessions. Surveys have shown that 70% to 75% of people plan to use some form of virtual care moving forward [6,7]. Analyses of outpatient claims data have also revealed that 20% of care could reasonably be virtualized. To sustain virtual care as we transition to a hybrid care model, we must reflect on the experiences of patients and clinicians in adopting virtual care technology, incorporate lessons from the digitization of health records, and identify ways to deliver intuitive and supported experiences.

Technology Adoption

Reflecting on recent clinic schedules, we wonder how the future of medicine can become digital by default. Although the uptake of virtual care before the COVID-19 pandemic had been modest, generally used by those comfortable with technology or seeking convenience, Inception Health, our innovation lab within the Froedtert & the Medical College of Wisconsin health network, had been laying the groundwork for broader digital health adoption. Reducing the complexity, cost, and hassle of health care were the goals, digital technology was the medium, and consumerism and competition were the levers. The first patient that either of us ever took care of through video-based care was a man in his 40s who worked in the information technology industry. He became a patient after presenting with diabetic ketoacidosis as an adult. After his diagnosis, he had diligently watched his diet and digitally tracked his blood glucose and he eventually stopped receiving any insulin-based therapy. He was an example of, as Everett Rogers, who framed the diffusion of innovation curve, would say, an “early adopter” [8]. He had comfort with the medium and the financial resources and capabilities to easily connect with a clinician. The COVID-19 pandemic and the limitations of in-person care have pulled Rogers’ diffusion of innovation curve leftward, dragging most patients and providers into a new mode of care. We were trying to get Ms J there, too.

And here lies one of the biggest challenges for digital care. Unless our profession achieves equal access and experiences

for all our patients, digital and video visits may amplify the divide in care quality between those able to easily access care and those who face major barriers: older or differently abled populations, underserved populations, and racial and ethnic minority populations [9]. Data have shown that up to 38% of older patients are unprepared for telemedicine [10]. Over one-quarter of Medicare beneficiaries lack any internet access altogether, an issue that disproportionately affects communities of color and patients with lower educational attainment [11]. Our experiences during the COVID-19 pandemic highlighted that minority patients were less likely to schedule video visits than telephone visits [12]. Furthermore, when patients did schedule video visits, we witnessed our older and more socioeconomically disadvantaged patients having lower success rates in connecting with their physicians [13].

Echoes of Electronic Health Record Adoption

As health care digital transformation is underway, much can be learned from the impact, pitfalls, and challenges of implementing EHR systems in the past decades that can inform us about the road ahead. As clinicians, we are already concerned that the EHR has turned us into data entry clerks, commandeering the practice of medicine and leading to “death by a thousand clicks” [14]. In parallel, this revolution of virtual care has begun to turn all medical staff into legions of technical supporters, adding to the list of required skills for certain jobs and the time required for visits. Pressing the “connect” button with each visit brings uncertainty. Will this work? Will we see and hear them, and will they see and hear us? A small amount of dopamine bursts when the patient’s image appears, coinciding with a sigh of relief.

For many patients, their telemedicine visits are their first virtual care experiences, and for many older physicians, EHRs may have been their first intensive computing experience. To connect to their clinicians, patients navigate instructions, download apps, and check themselves in, oftentimes with little assistance and using a mere 6-inch screen; they could experience difficulties at any step along the way. Ms J downloaded the required app, but she was still working through the check-in screens: “It says e-check-in, medications, allergies... but I don’t see what it wants me to do next.” Ms J had worked with our new digital support center after our initial discussion to ensure she installed the right mobile app and to log into her account, which required her to confirm her email account and set up a strong password with at least one special character. Our digital check-in configuration required our patients to verify their insurance, medications, and electronically sign any required documents, mirroring steps that staff usually take during in-person visits. The unfamiliar user interface, one that required scrolling to read small print on a mobile device, was difficult to overcome, and the user’s frustration was mounting. Despite our digital support center, medical assistants, and adaptation to the new role of digital physicians, we had failed to connect to the patient by video, a fate that became a pattern of a failure of digital care as a whole. In data from our organization’s experience, 1 in 10 scheduled video visits shifted to telephone visits [13].

As health care shifts to digital platforms, clinicians must learn from and avoid the mistakes we made during our attempts to

digitize medical records. Merely digitizing documents, adapting paper forms into electronic copies, accomplishes only a fraction of digital capability. Digital technology provides power by automating routine tasks, providing improved reliability and availability, and creating new possibilities. Although EHR implementations were a prerequisite for digital care, they created their own problems: they have been associated with increasing clinicians' workloads and physician burnout [15], were not easily interoperable with one another [16], contributed to increasing health care costs [17], became more complex over time [18], and were linked with a reduction in some elements of patient-physician communication [19]. A modern synopsis of EHRs indicates that they are often "feature rich, yet function poor" [20]. We may very likely amplify those challenges and pitfalls if they are not considered carefully as we transition to digital and virtual care services for patients.

Creating Better Experiences

To sustainably move forward, experiences like Ms J's, and many others, have taught us that we must take different approaches and consider technology as part of the fabric of care rather than a mere medium. At our organization, we have embarked on a reengineering and rebuild of our virtual care experience. Rather than trying to substitute video visits for in-person care, we should conceptualize how the continuum of the care experience can be redesigned, combining the advantages of virtual and in-person care, the power of computing with human empathy, and a seamless digital pathway with timely access to the care team. The following questions should be considered: Which patients scheduled for the day could be seen through quick digital or telephonic check-ins? How can we use digital technology to rightsize the attention needed rather than providing patients with a standard visit length?

The following 2 principles guide our approach for creating sustainable virtual experiences: (1) intuitive experiences, which aim to make digital care easily accessible across the technological literacy continuum without the need for extensive training and (2) digital navigation and support, which aims to reimagine support for patients as they navigate digitally native health care interactions.

Intuitive Experiences

Drs Warner Slack and Howard Bleich, 2 pioneering physician-informaticians, both often quipped that "the quality of the computing is inversely proportional to the thickness of the training material, or length of training" [21]. We must work toward building accessible experiences for patients that incorporate empathy for users and examine all steps along the user journey. This requires taking a 360-degree view of the end-to-end experience [22]. Steps to reduce the cognitive load experienced by users and create simple interfaces that focus on a single step or task at a time with minimal scrolling required may help [23]. At a minimum, we should start by questioning each step in the virtual visit chain, from app download to electronic check-in, that a patient is asked to complete: Is this truly required for clinical care, and how else may we accomplish it [24]?

Embracing agile principles, which promote outcomes rather than processes, may help sustain this flexibility and develop better ways of improving the value of the experience for patients [25]. Working closely and collaboratively with patients as users of the tools and using frameworks like human-centered design with frequent user-testing, can identify design flaws early and help create more intuitive experiences.

Digital Navigation and Support

The goodwill of clinicians to provide technical support, as one of us was doing that Friday evening, is a very scarce resource. Clinicians must take care of people, not their technology, or so we believe. "The tech just needs to work," a colleague told us. And yet, technology is far from the only issue. In clinical informatics, we often use the sociotechnical theory to guide technology implementations, which requires an understanding of people, processes, human-computer interactions, technology, and the interdependencies of these components [26]. Our implementation followed these tenets, but perhaps from the perspective of early adopters, not late adopters. Multiple layers of support are likely needed, such as having clinical staff who are knowledgeable about common issues, navigation support built into the digital experience, and family or community support for the technology.

Technical support may also become part of the care team, similar to how health coaches focus on assisting patients with behavior change or social workers support other needs such as transportation. A central competency of technology will be required across the caring professions; medical assistant training, for example, may have basic digital health technology as required learning. This know-how will also be crucial for encouraging patients to adopt other digital tools, such as digital therapeutics, that add evidence-based support in the form of apps or software [27].

In the hallways of our hospitals, staff with bright red blazers greet patients as they enter and help them navigate the hospital to reach their clinic appointments. What is the equivalent of such an experience in the digital realm? It may take new and different forms that health care systems must be willing to try. Perhaps avatars or bots may help patients prepare for visits, with the ability to connect with a support person if a patient has difficulty accessing their doctor. A successful digital support structure might become a standard in all health care systems rather than an afterthought.

For people who need more "at the elbow" support, or who lack internet connectivity or the ability to use technology, more instrumental support may be required. Partnerships with other organizations, such as those that already provide technical support as their core business, may also emerge to better support patients. For example, centralized support stations that can assist patients with telemedicine visits may open, either in satellite clinics, like more traditional telemedicine originating sites, or in community locations, such as pharmacies or senior housing.

What we have seen during the COVID-19 pandemic is that traditional ambulatory care in brick-and-mortar facilities plummeted, and virtual care was the primary option for nonemergent issues [28]. Virtual care was our main form of

care; over the span of a few weeks, its users grew from early adopters to nearly everyone else. And although we saw challenges, such as those outlined, we also saw heroic achievements. The patient who had a virtual visit during a smoke break to discuss quitting, the immunocompromised patient who was worried about a COVID-19 rash (it was shingles), and the patient with unstable transportation who was able to see us the same day all come to mind as incredible victories. At a systems level, we must do all that we can to focus on the care, not the medium, to heal and care for our communities.

As for Ms J, she scheduled an in-person visit after the weekend. She was able to send in a picture of her foot using her phone camera, a consolation victory for the effort in connecting her to our portal, allowing us to triage her care. At her visit, it

became clear, aided by the ability to attentively listen and observe, that her foot pain and the bumps were unrelated matters, and a diagnosis of plantar fasciitis and incidental varicosities was made, rather than a bug or spider bite. The only “bug” in the clinic was the difficulty in navigating the digital process.

Based on this lesson and others like it, our organization has implemented additional video tools to enable failover, circumventing barriers of portal sign-in and check-in requirements and adding more resources to our digital support arsenal. More broadly, we are scrutinizing every facet of the virtual care journey. We imagine others are, or should, be doing so as well.

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Conflicts of Interest

None declared.

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Abbreviations

EHR: electronic health record

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Viewpoint

Moving to Personalized Medicine Requires Personalized Health Plans

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Abstract

When individuals, families, and employers select health plans in the United States, they are typically only shown the financial structure of the plans and their provider networks. This variation in financial structure can lead patients to have health plans aligned with their financial needs, but not with their underlying nonfinancial preferences. Compounding the challenge is the fact that managed care organizations have historically used a combination of population-level budget impact models, cost-effectiveness analyses, medical necessity criteria, and current medical consensus to make coverage decisions. This approach to creating and presenting health plan options does not consider heterogeneity in patient and family preferences and values, as it treats populations as uniform. Similarly, it does not consider that there are some situations in which patients are price-insensitive. We seek to highlight the challenges posed by presenting health plans to patients in strictly financial terms, and to call for more consideration of nonfinancial patient preferences in the health plan design and selection process.

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KEYWORDS

quality-adjusted life years; health insurance; personalized outcomes; patient preferences; cost-effectiveness; managed care

Introduction

In the United States, there are many health plan designs in use. When individuals, families, and employers select plans, they are typically only shown the financial structure of the plans and their provider networks. This variation in financial structure can lead patients to have health plans aligned with their financial needs, but not with their underlying preferences—such as their desire for their health plan to cover or not cover family planning services. Compounding the challenge is the fact that managed care organizations have historically used a combination of population-level budget impact models, cost-effectiveness analyses, medical necessity criteria, and current medical consensus to make coverage decisions. This approach to creating and presenting health plan options does not consider heterogeneity in patient and family preferences and values, as it treats populations as uniform. Similarly, it does not consider that there are some situations in which patients are price-insensitive. [1] We seek to highlight the challenges posed by presenting health plans to patients in strictly financial terms,

and to call for more consideration of nonfinancial patient preferences in the health plan design and selection process.

Personalization and the Patient

Patients and their families can be directly involved in the process of valuing health plan attributes. Currently, there are several health plan decision support tools, such as Picwell and PLANselect, which help patients and families select health plans by answering questions about their financial preferences. The decision-making process is centered around answering questions related to premiums, deductibles, and other financial characteristics. Clinical questions typically relate to the anticipated frequency of health care utilization and prescription medications used. Once these questions are answered, a number of health plans are presented as options for patients and families, along with information about their financial characteristics (monthly premium costs, copays, and deductibles) and health care providers available in each plan's network. Given the information provided, it is not possible for patients and families to understand how the coverage they are being offered aligns

with their nonfinancial preferences and values. Information to consumers tends to lack transparency and details on the coverage of services for which preferences can vary owing to attitudinal differences, such as complementary and alternative medicine, medical abortion, or care at the end of life.

While current decision support tools simplify the health plan selection process for patients and their families, they ignore the underlying differences in the coverage policies between plans and may match an individual or family with a health plan whose coverage policies are not aligned with their values. The potential for misalignment between health plan coverage and personal values has been highlighted in the United States in the context of abortion, where in 2018, approximately half of US adults surveyed were found to support health plan coverage for abortion and approximately half did not support health plan coverage for it [2]. At present, it is so difficult to fully comprehend health plan details that even human resources departments, who typically make decisions regarding health plan benefit designs on behalf of a company or organization, are challenged. In one instance, the Catholic University of America inadvertently offered a health plan with limited abortion coverage before later discovering that their insurer had modified the plan's design without informing them [3]. Health plan decision support tools currently do not provide patients and families adequate support in assuring that the coverage policies of the health plan that they are selecting aligns with their beliefs, preferences, and values.

Today, a variety of methods, including standard gambles, time trade-offs, discrete choice conjoint analysis, and willingness to pay are used to elicit public preferences for health care services, with conclusions extrapolated to large and varied populations [4]. Going forward, a more tailored approach could be used in which patients and their families can be directly involved in the process of valuing financial and nonfinancial health plan attributes, and then paired with plans that align with their preferences individually rather than plans reflecting general societal norms that are only tailored on the basis of financial preferences. Moving toward a system in which patient and family preferences are better reflected in plan designs requires a redefining of the plan "shopping" experience. Rather than merely asking consumers whether they would wish to have a higher premium or a higher deductible, or whether they wish to have reduced premiums in exchange for reduced provider choice, health plans can additionally compete on the degree to which preferences over nonfinancial aspects of coverage (eg, the range and duration of health services covered and the provision of nonmedical services addressing the social determinants of health) are being satisfied.

The approach used to determine the services covered by a health plan may need to differ in an environment in which health plans are selected by individuals and employers (as is the case in the United States), rather than by a public payer (as is the case in the United Kingdom). Using quality-adjusted life years (QALYs) to facilitate decision-making related to health plan coverage—as a special task force of the Professional Society for Health Economics and Outcomes Research has recommended that US payers do—may not be fit for purpose if the health outcomes considered in calculating the QALYs

generated by an intervention are weighted uniformly for everyone living in the country [5]. The preferences of individuals and employers can vary greatly [6]. Research has also shown that the social value of an incremental QALY is not universal across individuals but instead depends on whether a person is nearing the end of life and may also depend on the person's prospective burden of illness [7].

Personalized cost-effectiveness analyses may be particularly valuable to people living with disabilities. While individual underwriting was banned in the United States by the Affordable Care Act, personalized cost-effectiveness analyses differ from underwriting in that they can be used to determine the benefits covered by the policy itself, rather than its pricing. Managed care organizations can—and do—offer a range of different health plans, at different pricing, with different attributes. The United States National Council on Disability has called for a moratorium on the use of QALYs in decision-making for Medicare and Medicaid (public health insurance programs) on the grounds that QALYs devalue interventions that extend the lives of people with disabilities and that mitigate the impact of disability on health [8]. Compared to interventions provided to people without disabilities, those provided to people with disabilities generate fewer future QALYs, thus driving discriminatory policies that may deprioritize people with disabilities [9]. This discrimination is exacerbated by the general practice of having people without disabilities participate in the assignment of QALYs, as people without a particular disability rate their expected quality of life with the disability as lower than do people living with that disability [10]. By engaging people with disabilities in the process of designing policies for similarly situated individuals by proactively seeking information on their preferences, plans can develop benefits that are better aligned with the people they serve.

Personalization and the Health Plan

As we approach an era of personalized medicine, we may need to enter an era of personalized health plans, in which patients and employers can choose from among a wider variety of health plans that differ in both their financial structures and the values that they capture. These include, for instance, the following:

- Should a health plan seek to extend life at all costs?
- Should a health plan offer coverage for services that may shorten or end life?
- Should a health plan cover services that prioritize convenience over quality?
- Should a health plan allow patients the flexibility to choose their provider even if their preferences may lead them to seek high-cost or low-quality providers?
- Should a health plan offer coverage for alternative therapies backed by minimal scientific evidence?

There is no single correct answer to these questions, but in a system in which there is a degree of consumer and employer choice, people and employers can potentially be paired with preference-aligned plans.

Conducting single cost-effectiveness analyses for interventions does not enable payers to adequately deal with the diversity of

the patients who they serve. QALYs seek to express the value of changes in quality and length of life in a single metric, and have become a widely used measure of health benefits in cost-effectiveness evaluations [11]. Wrapped up in the logic of the QALY is the premise that a payer will be willing to cover “cost-effective” therapies, and that the QALY gains from a given intervention are assumed to be of the same value, irrespective of the preferences and nonhealth characteristics of the patients [12]. We already recognize, however, that there are some differences among populations, as countries have assigned different weightings to the health outcomes used to compute a QALY, as well as different implied monetary values to a QALY, and thus an intervention with the same costs could be considered cost-effective in one jurisdiction but not in another [13]. Similarly, different individuals have different preferences, and thus there is scope for greater patient participation in determining what is cost-effective for each group or individual rather than simply for society as a whole.

The growth of personalized medicine also raises challenges to the generalizability of QALYs [14]. While a particular small-molecule treatment may be used to address multiple indications, all patients are ultimately taking the same drug. In contrast, when patients access and use a given digital therapeutic for different indications, they may be receiving different interventions, which aim to address different health concerns. For instance, a single app may offer a fully self-guided treatment to people with mild depression but a more expensive, therapist-guided intervention to people with moderate depression [15]. Although the specific app itself is the same in both cases, the treatment it provides and the cost of delivering that treatment varies in accordance with the indication for which it is used. Likewise, the number of QALYs generated by the app vary in accordance with how it is used. Generalizability issues are likely to extend into other forms of treatment as well because personalized medicine increasingly leads to the tailoring of

biological and chemical interventions, in place of the traditional “off-the-shelf” treatments used previously.

The standard QALY approach is based on eliciting of the preferences of members of the public over different health outcomes, where the strength of preference is determined by trade-offs against life expectancy or risk of death. There are serious problems with the ability of such preferences to serve as good guides to the relative impact of different health outcomes on peoples’ lives [16]. A more robust and reliable approach might therefore be to conduct assessments that allow peoples’ reports of their well-being and values to be used to determine the relative weights allocated to different health outcomes [17]. By developing a menu of different health plan offerings with different weights, payers can enable patients, families, and employers to more readily select plans that fit their preferences and values, with the understanding that not everyone would assign the same weights to outcomes when determining the QALYs experienced in a health state.

Conclusions

Irrespective of the details of valuing the outcomes produced by medical interventions, more patient participation in determining health plan coverage decisions will become necessary, especially as personalized medicine is playing an ever-increasing role in care. For more accurate assessments of the utility produced by interventions covered by health plans to be made, patients must more actively share their own preferences so that they may be properly accounted for in the decision-making model. Payers should work with patients and employers toward developing health plan population-specific coverage decisions more consistent with the preferences of the populations that these plans serve. Ultimately, personalizing medicine will require a new, more tailored approach to determine the health services that health plans should cover.

Authors' Contributions

PD conceptualized the study. AP and PD performed the literature search. AP drafted the manuscript. AP and PD edited the final draft of the manuscript.

Conflicts of Interest

None declared.

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Abbreviations

QALY: quality-adjusted life year

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Viewpoint

A Health Equity–Oriented Research Agenda Requires Comprehensive Community Engagement

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Abstract

Health policy and research communities have taken new approaches to addressing health equity, going beyond traditional methods that often excluded the contributions of health care consumers and persons with lived experience. This reevaluation has the potential to drive critical improvements in how we conduct research and innovate policy toward reducing health and health care disparities in the United States. Such considerations have led Fountain House, the founder of the Clubhouse model for peer-based psychosocial rehabilitation for persons with histories of serious mental illness, to incorporate community-based participatory action research (CBPAR) protocols within their research and service programs. The combination of CBPAR research methods within novel participatory care settings like Clubhouse programs presents unique and informative opportunities for the advancement of innovative health equity approaches to consumer empowerment in health care. In this piece, the authors (two staff researchers and one member researcher) propose how CBPAR research methods conducted in Clubhouses can uniquely advance equity-focused research methods, and how the benefit and enhancements from equity-focused research are continuously applied, practiced, and accountable to the communities within which the research is conducted. Embedding CBPAR practices within participatory care settings like Clubhouses, creates novel opportunities for research work to not only become more equitable but also become a part of the rehabilitative process, empowering the main beneficiaries of the research with the means to sustain and achieve further improvements for themselves. Such experiences are particularly important within rehabilitation settings, where there is a process of reclaiming empowerment and self-efficacy over a disability or illness and the social circumstances surrounding those conditions. Different stakeholders can all play important roles in advancing health equity-oriented research agendas by leveraging CBPAR principles. Academics and others in the research community can more comprehensively embed CBPAR methods into the design of their research studies. A critical link exists among how researchers conduct their studies, how providers organize care delivery and support, and how health plans pay for and evaluate care. CBPAR-generated research needs to fully engage clinical teams to ensure that ongoing community-involved care settings have direct applications to real-world care delivery. It is equally important that providers fully engage with their communities as they adjust their approaches to supporting the populations they serve.

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KEYWORDS

mental health; community-based participatory action research; health equity; serious mental illness; health policy; research; community; engagement; disparity; participatory care; consumers

Introduction

In recent years, health policy and research communities have adapted and rethought traditional approaches to health equity that often excluded the contributions of health care consumers

and persons with lived experience. This reevaluation has the potential to drive critical improvements in how we conduct research and innovate policy toward reducing health and health care disparities in the United States, consistent with the recent 2021 call to action by the Robert Wood Johnson Foundation (RWJF) “for its health equity agenda, the Biden administration

needs research that focuses on impacted communities” [1]. Moving this equity agenda forward requires providers and researchers to fully embrace participatory strategies with the communities they serve in both the conduct of participatory research and in the development of participatory care environments, where research benefits can persist within.

Such considerations have led Fountain House, the founder of the Clubhouse model for peer-based psychosocial rehabilitation for persons with histories of serious mental illness (SMI), to incorporate community-based participatory action research (CBPAR) protocols within their research and service programs. Central to the Clubhouse model is the joint operation of its services by professional staff working side by side with Clubhouse members (people with an SMI who join the Clubhouse have always been called members rather than patients or clients) in all aspects of Clubhouse program operations. Clubhouses intentionally structure therapeutic experiences and growth through the shared work of Clubhouse programs, emphasizing socialization and member empowerment to combat loneliness and stigma while also connecting members to traditional health and social support services.

Recognizing the synergy between CBPAR research methods with participatory care settings like Clubhouse programs presents unique and informative approaches to the advancement of health equity-focused research that involves consumer empowerment and continuous participation. In this piece, the authors (two staff researchers and one member researcher) propose how CBPAR research methods conducted in Clubhouses can uniquely advance equity-focused research and how the benefit and enhancements from this research are continuously applied, practiced, and accountable to the communities within which the research is conducted.

Defining CBPAR

CBPAR starts with the principle that all aspects of research should involve true collaboration among professional researchers and community of interest stakeholders, or colloquially, it holds true to the mantra “nothing about me without me.” Various formal definitions and approaches of CBPAR and community-based participatory research (CBPR) have been advanced. We embrace the same definition as used in the Chicago Health Disparities Study adapted from the WK Kellogg Foundation’s Community Health Scholars: “CBPR is a collaborative approach that involves all partners in the research process. [It] begins with a research topic of importance to the community...[combining] knowledge and action for social change to improve communities and eliminate disparities” [2].

Going beyond aligning research with community priorities and experiences, CBPAR methods also prioritize the training of community participants in scientific design and procedures so that the community can collaborate in research decision-making from a shared knowledge position. This level of participation and training empowers the community to leverage skills for continued assessment and advancement of the community’s interests beyond the scope and limitations of a given research study [3]. This is specifically relevant for the “action” processes of CBPAR, where research developed toward change-oriented

solutions can be implemented, sustained, and enhanced on an ongoing basis within the communities where the research was conducted.

CBPAR methods have particular importance for communities of interest that have historically been marginalized from participating in larger social systems that impact their daily lives. Some prominent CBPAR practice examples have occurred within indigenous communities managing diabetes prevention resources [4], migrant communities accessing social service resources [5], and mental health communities seeking greater advocacy for addressing social determinants of health related to poor outcomes [6]. Rather than being a burden, the empowering benefits of such collaborative approaches often enhance research quality, demonstrating more realistic and practical results due to the introspective data and action-oriented decision-making provided by community stakeholders in research procedures [7,8].

Health Equity and CBPAR

Health equity has been defined in multiple ways. Borrowing from RWJF, “health equity means that everyone has a fair and just opportunity to be as healthy as possible.” The RWJF definition further elaborates that health equity “requires removing obstacles to health such as poverty, discrimination, and their consequences, including powerlessness and lack of access to good jobs with fair pay, quality education and housing, safe environments, and health care.” Powerlessness and access barriers, in particular, have been parallel obstacles to equity in both health and health research [9].

In alignment with health equity goals, CBPAR methods offer unique research strategies to help address larger systemic issues related to health care accessibility, health literacy, and poor patient experiences [10]. However, certain change-oriented CBPAR outcomes can be complicated in many traditional public health settings, where imbalanced provider-patient power dynamics persist in terms of care decision-making, priority, and quality [11]. This power imbalance is particularly salient for persons with histories of SMI who can be forced to receive compulsory care within settings from which they are often disenfranchised.

Given such circumstances, while CBPAR methods can be readily used in traditional health settings, the persistent empowerment of patient communities to participate in the continuous change-oriented enhancements, delivery, and assessments of their own ongoing care is often limited [12]. To overcome these limitations, CBPAR and health equity agendas should pursue greater applications within a broader community-oriented approach to health care delivery that incorporates participatory practices in their core service model.

The Clubhouse Model: Maximizing Participatory Potential for Health Equity

Although minimal in their overall presence in health care, there are some rehabilitation settings that operate unique care models focused on uplifting consumers into roles of treatment

decision-making and peer-support delivery. A historical leader in such approaches is the Clubhouse model, a community-based psychosocial rehabilitation program for persons with histories of SMI. Founded in 1948 by persons with an SMI, the New York City-based Fountain House launched the Clubhouse movement with the purpose of creating communities of lived experience, where persons with histories of SMI could support and care for one another in their recovery journey.

Clubhouse programs offer strength-based peer interventions to help persons with SMI socially reintegrate and achieve agency in their health, quality of life, and care. This is achieved through the creation of an intentional peer community, where members are invited to (co)operate and administer Clubhouse operations, working side by side with Clubhouse professional staff to either receive or provide a range of social support services [13]. Member participation in Clubhouse services occurs through a structured work-ordered day, where members participate in the administration and delivery of peer-based support programs that often include education, care management, research, wellness, employment, and housing [14]. These facilitated experiences of shared contribution and administration drive what the Clubhouse calls the need to be needed, rehabilitating member agency, self-confidence, skills, and social acceptance, which have often been disrupted by shared histories of disenfranchisement, stigma, and diminished quality of life opportunities [15]. At the core of the Clubhouse model, every program decision, activity, and service offering involves member contributions, decision-making, and administration to the benefit of not only the consumers of Clubhouse services but also the member stakeholders who jointly run the Clubhouse alongside professional staff.

Recognizing the participatory congruence between Clubhouse model and CBPAR research methods, Fountain House has sustained a legacy of incorporating CBPAR practices in its research initiatives. Programmatically, this has taken the form of Fountain House maintaining a longstanding Research Unit as one of its program service areas, where members learn and direct the community's research interests and priorities. Members and staff have also created a Research Committee to manage high-level administrative decisions in developing research collaborations across the national Clubhouse network and public health policy agenda. These continuous peer-led research forums empower members to develop research skills, translate their research priorities, and self-administer the change-oriented outcomes of their research toward positive program enhancements in their own care settings. The opportunities, insights, and skills developed through CBPAR within peer-driven programs like Clubhouse allows not just for health equity research advancements to be discovered but for them to also be accountably enacted and implemented by the very people whom they are intended to benefit within the settings they help operate.

One example of this unique CBPAR health equity dynamic within Clubhouse care settings occurred in a collaboration between Fountain House and Yale University, where Clubhouse members were trained in qualitative research to conduct an analysis of member care experiences, trajectories, and differing needs within the community. The results of this study, conducted

from start to finish by members, informed programming decisions around new member orientation and needs assessment procedures that seek to engage members during "critical periods" of early membership, identifying a spectrum of member experiences interacting with the Clubhouse as either a supportive stepping stone or a long-term community destination. The members who administered the study were able to inform new program practices and further apply their research training toward training other members and even work as paid research consultants and coders in future research collaborations. This has been the case with a current CBPAR project with Harvard Medical School to co-design a virtual healthy lifestyle intervention that seeks to involve members not only in the development and implementation of the research study but also in administering components of the intervention themselves, after the study's completion. What we have seen from this approach with Harvard, in addition to other CBPAR projects, is that members of different racial, social, and health backgrounds actively engage and adjust interventions and research protocols that address their collective needs, thus driving more equitable care support approaches that they can supervise and perform continuously.

By embedding CBPAR practices within participatory care settings like Clubhouses, the research work not only becomes more equitable but also restorative, empowering the individuals who are meant to benefit from the research with the means to achieve that improvement for themselves. This is especially important within rehabilitation settings, whereby in virtue of being in need of rehabilitation, there is a drive to reclaim empowerment and self-efficacy not only over a disability or illness but also the social circumstances surrounding those conditions. This has been the experience of one of the authors, who took an academic leave for mental health reasons, engaged with the CBPAR program at Fountain House, and is now undertaking academic pursuits toward developing strategies for consumer-informed solutions within the mental health industry.

Moving Forward: Advancing a Health Equity Research Agenda

Different stakeholders can all play important roles in advancing health equity-oriented research agendas by leveraging CBPAR principles. Academics and others in the research community can more comprehensively embed CBPAR methods into the design of their research studies. Although the National Institutes of Health has a CBPAR program in its National Institute on Minority Health and Health Disparities, it has not built CBPAR requirements into its standard grantmaking process. National Institutes of Health and other federal agencies can create more robust expectations for applying researchers and support the promotion of community health settings, where CBPAR practices can be fully enacted for the continued benefit and empowerment of patient communities and their role in care design and delivery.

How researchers conduct their studies will benefit from greater coordination with and application within participatory care settings. To further enhance health equity-focused research approaches, CBPAR-generated research needs to fully engage

clinical teams and consumer communities to ensure that ongoing community-involved care settings have direct applications to real-world care delivery. This is particularly important for exploring and promoting innovations in how health plans pay for certain types of care. Indeed, providers are unlikely to shift their models unless health plans and other payers embed community-based participation and human-centered design into their payment models. The final piece of this effort relates to the role that state and federal policy making has on this intersection between health equity and community-oriented

research models. Policy makers—those involved in both legislative and regulatory aspects—need to fully embrace CBPAR as one of several vehicles for advancing a national agenda to promote health equity, which includes the investment and promotion of participatory and peer-driven treatment settings where CBPAR methods can be implemented. Ultimately, how we measure and improve health equity will be dramatically influenced by the research questions we ask and the way we conduct that research. The communities we want to support must be integrally involved.

Conflicts of Interest

None declared.

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Abbreviations**CBPAR:** community-based participatory action research**CBPR:** community-based participatory research**RWJF:** Robert Wood Johnson Foundation**SMI:** serious mental illness

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Original Paper

Evaluating a Global Assessment Measure Created by Standardized Patients for the Multiple Mini Interview in Medical School Admissions: Mixed Methods Study

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Abstract

Background: Standardized patients (SPs) are essential stakeholders in the multiple mini interviews (MMIs) that are increasingly used to assess medical school applicants' interpersonal skills. However, there is little evidence for their inclusion in the development of instruments.

Objective: This study aimed to describe the process and evaluate the impact of having SPs co-design and cocreate a global measurement question that assesses medical school applicants' readiness for medical school and acceptance status.

Methods: This study used an exploratory, sequential, and mixed methods study design. First, we evaluated the initial MMI program and determined the next quality improvement steps. Second, we held a collaborative workshop with SPs to codevelop the assessment question and response options. Third, we evaluated the created question and the additional MMI rubric items through statistical tests based on 1084 applicants' data from 3 cohorts of applicants starting in the 2018-2019 academic year. The internal reliability of the MMI was measured using a Cronbach α test, and its prediction of admission status was tested using a forward stepwise binary logistic regression.

Results: Program evaluation indicated the need for an additional quantitative question to assess applicant readiness for medical school. In total, 3 simulation specialists, 2 researchers, and 21 SPs participated in a workshop leading to a final global assessment question and responses. The Cronbach α 's were >0.8 overall and in each cohort year. The final stepwise logistic model for all cohorts combined was statistically significant ($P < .001$), explained 9.2% (R^2) of the variance in acceptance status, and correctly classified 65.5% (637/972) of cases. The final model consisted of 3 variables: empathy, rank of readiness, and opening the encounter.

Conclusions: The collaborative nature of this project between stakeholders, including nonacademics and researchers, was vital for the success of this project. The SP-created question had a significant impact on the final model predicting acceptance to

medical school. This finding indicates that SPs bring a critical perspective that can improve the process of evaluating medical school applicants.

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KEYWORDS

co-design; participatory design; medical schools; exploratory sequential mixed methods design; school admission criteria; medical students; communication; multiple mini interviews; interview; patient; student; medical school; acceptance; study design

Introduction

People trained to act out a role consistently and repeatedly in a realistic way for active learning and assessment purposes in medical education are called standardized patients (SPs) or simulated participants [1,2]. They will not only act in the simulations but also assist in the assessment of learners. Ample evidence suggests that SPs can effectively assess and evaluate medical students, nursing students, medical residents, and other clinical learners [3-5]. They are essential stakeholders and collaborators within the medical education process who help learners at all levels meet educational objectives [1,2,6].

At our institution, our team of SPs partners with the simulation center staff to create a safe environment for learners to fail and make mistakes. The practice of medicine is complex and nuanced, which requires physicians to have the skills and abilities to work cross-culturally with patients and families to make life and death decisions every day. It is difficult to learn these needed skills simply in a classroom; practical and practiced application of the assessable skills within a simulated environment is needed. The simulated environment must not only be a place to learn and practice the skills and abilities but also to fail and make mistakes. This safe and simulated environment is the fundamental link to the education of compassionate and competent future physicians.

Simulation center demonstrations and recruitment events, as well as referrals by current SPs or staff members, allow for the incorporation of new SPs at our institution. People who become part of the simulation center team as SPs come from all walks of life and represent various demographics, work histories, and experiences. All SPs are trained to recreate the history, personality, physical findings, emotional structure, and response pattern of an actual patient accurately and consistently during a simulated experience or scenario. They are also trained to assess learner performance and provide individualized feedback to learners in a constructive manner. After training, the SPs partner with staff in delivering the medical scenarios to learners where the objectives focus on skill development. Equally important are nonmedical scenarios that focus on the development of verbal and nonverbal communication skills. In the nonmedical scenarios, they teach students through practical application, assessment, and active learning. Thereby, SPs along with the rest of the medical school faculty and staff coeducate

and codevelop these learners into future physicians. Additionally, they not only help learners during their educational experiences but are also core team members during the medical school admissions process [3,7].

Many medical schools have moved toward a holistic admissions process. This process can include the assessment of applicants' noncognitive skills such as moral reasoning and interpersonal communication through the inclusion of multiple mini interviews (MMIs) [7-17]. MMIs are designed to evaluate the applicants' listening abilities, professionalism, ethics, empathy, integrity, cultural sensitivity/humility, problem-solving skills, and communication skills [9,10,15,18,19]. During the MMI, each applicant rotates through a series of stations where SPs evaluate their noncognitive traits [7,8,12,20-22]. SPs have been identified as a valid and reliable resource to evaluate applicants in an MMI process [3,23].

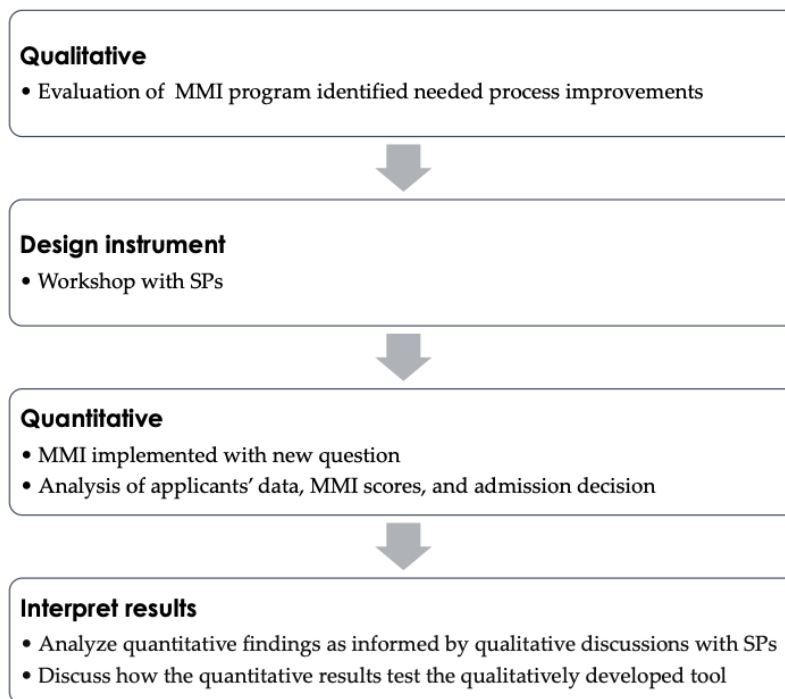
The inclusion of stakeholders in the design and implementation of interventions that impact their own population can provide more robust outcomes [24-29]. Additionally, meaningfully involving patients and the public in the co-design of interventions produces studies that are more patient-centered, less disruptive for study participants, and more accepted and valued by the study population [30-36]. The literature has shown that when SPs codevelop simulation scenarios and educational experiences, they can enhance outcomes for learners and educators [37-39]. However, we found no evidence for the inclusion of SPs in the process and development of an evaluation instrument, tool, or rubric for use during simulated interactions with learners or medical school applicants. Therefore, the purpose of this study was to describe the process and evaluate the impact of having SPs cocreate a global measurement question that assesses medical school applicants' readiness for medical school.

Methods

Study Design

This study followed a participatory, exploratory, sequential, and mixed methods design (Figure 1) [40-43]. Exploratory designs are commonly used for developing new instruments or measures [43-45] and are particularly useful when a culturally responsive instrument needs to be created [43].

Figure 1. Illustration of participatory, exploratory, sequential, and mixed methods design flow. MMI: multiple mini interview; SP: standardized patient.

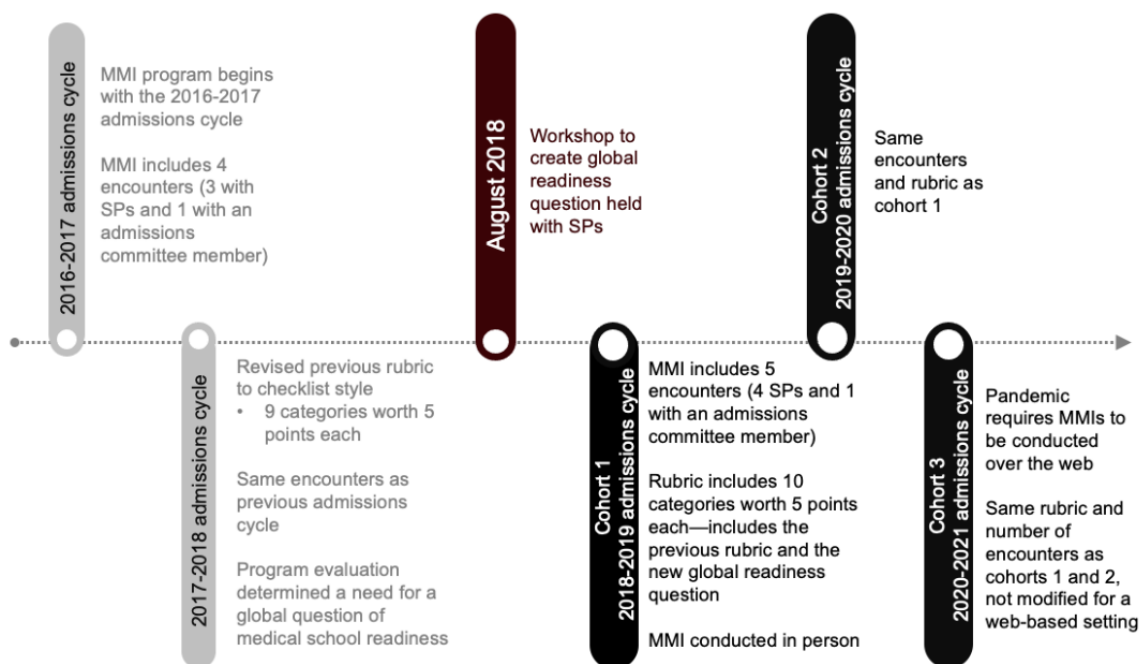


The MMI Program

The original MMI process was implemented in the 2016-2017 admissions cycle [15], and a timeline of the program procedures and revisions can be found in Figure 2. The MMI encounters that are a part of our admissions process are delivered and evaluated by our simulations center team which includes simulations specialists and SPs. These encounters require no medical knowledge by the applicant and can simulate communication with a patient, peer, or coworker. For each MMI

encounter, the applicant interacts with a single SP while being observed through a 2-way window by another independent SP in real time. At the conclusion of the encounter, the observing SP and performing SP collaboratively discuss the applicant’s performance and skills and complete the rubric to provide a quantitative outcome. The evaluation by 2 SPs helps eliminate potential bias, ensures accuracy in scoring, and streamlines the fast pace of MMIs. Furthermore, a standardized rubric and 2 SPs ensure accuracy and timely completion in the limited time allotted between encounters.

Figure 2. Timeline of the MMI program and evaluation beginning in 2016 through program evaluation and 3 cohorts for analysis. MMI: multiple mini interview; SP: standardized patient.



MMI Program Evaluation

SPs, simulation center staff, and members of the admissions committee participated in an evaluation of the MMI program after the 2017-2018 cycle. This evaluation included a review of the MMI rubric and admissions committee documents for quality improvement purposes. According to evaluation results, partnering with SPs to cocreate an addition to the evaluation rubric could improve the MMI program.

SP Workshop

For the SPs involved in the MMI during the medical school admissions process, the simulation specialists and researchers hosted a workshop in August 2018 to allow for the cocreation and improvement of the rubric. The workshop included a short didactic session and active participation by the SPs. During the didactic presentation ([Multimedia Appendix 1](#)), the researchers gave a short presentation to (1) review the rubric used in the previous year, (2) present reasons for creating the new question, (3) explain the basic survey design [46], and (4) discuss the importance of stakeholder engagement in this type of work.

Upon completing the presentation, the SPs were divided into small groups and given approximately 30 minutes to create 1 question and the corresponding responses. The created questions were written and displayed on large pieces of paper placed on the walls and on 2 screens in the room. Subsequently, everyone was reassembled into a large group to discuss the developed questions. Voting and further discussion led to a single SP-created question. Once the question had been finalized, the SPs were then divided into 2 groups to write response options for the newly created question. Collaborative discussion, voting, and editing led to a co-designed set of response options.

Quantitative Data Collection and Analysis

The quantitative data were collected from various sources, including 1084 medical school applications, MMI process forms, and admission status for 3 cohorts: (1) 2018-2019, (2) 2019-2020, and (3) 2020-2021. Applicants' demographics were gathered from school applications. Averages of the 4 SP encounters for each MMI category were provided for individual applicants. Admission status was categorized into 3 categories: accepted, declined, or wait-listed (reviewed applicants awaiting consideration from those who withdrew their application prior to acceptance). The wait-listed applicants were excluded from the regression models. In all, 3 applicants did not have admissions or matriculations status listed and were excluded from analysis.

Statistical Analysis

Quantitative data analyses were performed using SPSS software (version 26; IBM Corp). Descriptive statistics included frequencies, means and SDs, and percentages. The internal consistency reliability of the MMI was measured using a Cronbach α test evaluating all 10 variables within the MMI: (1) opening the encounter, (2) empathy, (3) nonverbal behavior, (4) verbal behavior, (5) listens well, (6) therapeutic relationship, (7) negotiation of the plan, (8) closing the encounter, (9) rank of readiness, and (10) admissions committee member. To determine if the SPs may be influenced by the demographics of the applicants, the applicants' age, sex, and underrepresented

in medicine (URM) status (dichotomous yes/no) were regressed (multiple linear regression) on the average MMI score (excluding the admissions committee member score). Finally, we sought to determine if medical school admissions status could be predicted based upon the variables within the SP-scored MMI (9 variables). A forward stepwise binary logistic regression was used to identify possible predictors of acceptance status (accepted or declined) out of the following MMI candidate variables: opening the encounter, empathy, nonverbal behavior, verbal behavior, listens well, therapeutic relationship, negotiation of the plan, closing the encounter, and rank of readiness. The forward logistic regression used the likelihood-ratio test to enter or remove variables from the model.

Ethics Approval

Ethical oversight of the project was conducted by the Institutional Review Board at the University of South Carolina (Pro00069266).

Results

MMI Program Evaluation

Feedback from the SPs, simulation center staff, and admissions department revealed that the checklist rubric ([Multimedia Appendix 2](#)) was easy for SPs to use and for the admissions committee to integrate into their decision-making process. In contrast, some SPs would write extensive qualitative comments about applicants, whereas others would leave few or no notes on those they interviewed. Additionally, it was revealed that the admissions committee was not using these written comments by the SPs in their applicant deliberations. Therefore, adding a single MMI rubric item that evaluates applicants based on their interactions with SPs during the scenario might reduce the SPs' need to write qualitative comments and provide additional information for the admissions committee. To this end, a workshop was planned and implemented to develop a question and responses to provide a global assessment of a medical school applicant.

SP Workshop and Global Measure Development

In attendance at the workshop were 3 simulation specialists, 2 researchers, and 21 SPs. The mean age of the SPs was 53.1 (SD 12.43; range 27-70) years, and a majority were female (16/21, 76%) and had a bachelor's degree or higher (18/21, 86%). The 6 SP-created questions are listed in [Table 1](#). The SPs identified that 3 of the 6 questions were dichotomous in nature and concluded that if selected, these 3 questions would not be a useful global measure assessment tool. Additionally, they identified certain words that they deemed to be either useful or problematic, such as "successful," "characteristics," "1st year" (vs medical student in general), "ability," and "readiness." Several SPs deliberated over the ways to strengthen the questions. For example, the SPs suggested adding "Based on this interaction," to the beginning of the second question to help the person answering the question focus on the interaction with the applicant and not the applicant's qualities overall. Additionally, the question of whether communication skills could be developed or if they are innate and unchangeable was discussed.

At the end of the discussion, voting resulted in questions 6 and 4 receiving 10 and 9 votes, respectively (2 of the 21 SPs had to leave the workshop before the voting process). Additional discussion led to a “tie-breaking vote,” yielding question 6 as the top choice. The SPs, however, determined that both questions had some aspects they wanted to emphasize. Thus, a single question was merged from both, which resulted in the combined global question: “Based on the candidate’s communication and interpersonal skills, rate this candidate’s readiness for medical school.”

As shown in Table 2, both groups developed response options for the global readiness question. There was a brief discussion before a vote was taken, and rubric 2 was chosen with some minor group edits. The bottom 2 scale items were modified to add *readiness* in light of the question. The word *adequate* was replaced by the word *proficient* in the third scale item. On the *advanced* scale item, the words following the slash may be listed interchangeably instead of in the specific order listed.

Table 1. Original questions created by standardized patients.

Group number	Question
1	Based on this interaction, how successful do you think this applicant is likely to be as a medical student?
2	Will this candidate be a successful 1st year medical student?
3	Do you think this student demonstrates the characteristics of a successful medical student?
4	How well did this candidate demonstrate communication and interpersonal skills which will allow him/her to succeed in medical school?
5	Do you think the applicant will be a successful medical student?
6	Rate this candidate’s readiness for medical school.

Table 2. Standardized patient–developed response options for the developed readiness question.

Ranking score	Response options set 1	Response options set 2	Final response options set
Rank of 5	Exemplary	Exceptional/extraordinary readiness	Exceptional/extraordinary readiness
Rank of 4	Ready with minor concerns	Advanced/strong and engaged	Advanced/strong and engaged
Rank of 3	Ready with some concerns	Adequate/addresses basics	Proficient/addresses basics
Rank of 2	Serious concerns	Minimal	Minimal readiness
Rank of 1	Not ready	Did not demonstrate	Did not demonstrate readiness

Quantitative Results

Medical School Applicants Characteristics

Table 3 summarizes the characteristics of the 1084 medical school applicants who were selected and invited for an interview by the 3 corresponding cohort years and as a whole. Overall, a little over half (589/1084, 54.3%) of the interviewees were female, and their average age was 24.7 (SD 2.87) years. Of the 1084 interviewees, 20.1% (n=218) were from racial/ethnic communities considered as URM, and 12.5% (n=135) identified as African American or Black. In addition to the interviewees’ characteristics, Table 3 presents the average score and SD of

each of the MMI categories for each of the 3 cohorts. The average MMI score for rank of readiness (ie, the SP-created question: “Based on the candidate’s communication and interpersonal skills, rate this candidate’s readiness for medical school”) increased over time from 3.38 (SD 0.53) to 3.52 (SD 0.48) out of 5, with 5 being exceptional or extraordinary readiness. In contrast, the MMI score of the admissions committee member (ie, painting/image discussion) decreased over time from 3.64 (SD 0.92) to 3.45 (SD 0.82). Of those interviewed, 52% (199/383), 51.9% (187/360), and 60.7% (207/341) were offered admissions to the medical school in cohorts 2018-2019, 2019-2020, and 2020-2021, respectively.

Table 3. Descriptive statistics of interviewees by cohort year.

Variable	2018-2019 (n=383)	2019-2020 (n=360)	2020-2021 (n=341)	Total (N=1084)
Sex, n (%)				
Female	201 (52.5)	192 (53.3)	196 (57.5)	589 (54.3)
Male	182 (47.5)	166 (46.1)	145 (42.5)	493 (45.5)
Prefer not to answer	0 (0)	— ^a	0 (0)	—
Age (years), mean (SD)	25.44 (2.95)	24.92 (2.62)	23.68 (2.74)	24.71 (2.87)
Underrepresented in medicine, n (%)				
Total	57 (14.9)	73 (20.3)	88 (25.8)	218 (20.1)
African American or Black	32 (8.4)	43 (11.9)	60 (17.6)	135 (12.5)
American Indian or Alaska Native	—	—	—	8 (0.7)
Hispanic or Latinx	22 (5.7)	27 (7.5)	23 (6.7)	72 (6.6)
Multiracial or mixed race	—	—	—	—
Opening the encounter, mean (SD)	4.13 (0.60)	4.28 (0.61)	4.18 (0.56)	4.19 (0.59)
Empathy, mean (SD)	3.85 (0.75)	3.93 (0.76)	4.12 (0.61)	3.96 (0.72)
Nonverbal behavior, mean (SD)	4.57 (0.44)	4.49 (0.56)	4.24 (0.63)	4.44 (0.56)
Verbal behavior, mean (SD)	3.97 (0.44)	3.93 (0.62)	4.10 (0.50)	4.00 (0.57)
Listens well, mean (SD)	3.69 (0.51)	3.71 (0.51)	4.02 (0.50)	3.80 (0.53)
Therapeutic relationship, mean (SD)	4.30 (0.69)	4.24 (0.73)	4.35 (0.61)	4.30 (0.68)
Negotiation of the plan, mean (SD)	2.03 (0.64)	2.14 (0.62)	2.75 (0.68)	2.30 (0.68)
Closing the encounter, mean (SD)	3.46 (0.63)	3.54 (0.66)	3.71 (0.57)	3.56 (0.63)
Rank of readiness, mean (SD)	3.38 (0.53)	3.46 (0.57)	3.52 (0.48)	3.45 (0.53)
Admissions committee member score (painting/image discussion), mean (SD)	3.64 (0.92)	3.36 (0.92)	3.45 (0.82)	3.48 (0.90)
Acceptance status, n (%)				
Accepted	199 (52)	187 (51.9)	207 (60.7)	593 (54.7)
Declined	122 (31.9)	146 (40.6)	111 (32.6)	379 (35)
Wait-listed	62 (16.2)	27 (7.5)	23 (6.7)	112 (10.3)

^aCells with n≤5 were suppressed to protect the identity of the individuals.

Evaluation of MMI

The 10-variable MMI had a high level of internal consistency overall and in each cohort year as determined by Cronbach α 's of 0.877 (all years combined), 0.89 (2018-2019), 0.90 (2019-2020), and 0.87 (2020-2021).

Regression Models for MMI Scores

We ran a multiple regression to predict the average MMI score from age, sex, and URM status. The multiple regression model for all cohorts predicted the average MMI score ($P<.001$, $R^2=.035$). Of the 3 variables, 2 (sex and URM status) added statistical significance to the prediction ($P<.001$ and $P=.003$, respectively). The results of the multiple regression models of the individual cohorts can be found in [Table 4](#).

Although MMI average scores are used for admission committee discussions, this analysis is aimed at determining how much impact each of the variables within the MMI may have on the acceptance decision—specifically, to determine if the readiness for medical school should be selected as a predictor variable. A forward stepwise binomial logistic regression was performed to ascertain the effects of the MMI variables (opening the encounter, empathy, nonverbal behavior, verbal behavior, listens well, therapeutic relationship, negotiation of the plan, closing the encounter, and rank of readiness) on the likelihood that applicants would be accepted to medical school. Models were built for all cohorts combined and each cohort individually. [Table 5](#) contains results from all groups and models.

Table 4. Multiple regression results for MMI average.

MMI ^a average	Unstandardized coefficient			Standardized coefficient (β)	R^2	ΔR^2	P value
	B	95% CI	SE				
2018-2019 cohort model					0.08	0.07	<.001
Constant	2.63	2.19-3.06	0.22	N/A ^b			<.001
Sex ^c	0.16	0.07-0.25	0.05	0.17			<.001
Age	0.03	0.02-0.05	0.01	0.22			<.001
URM ^d status	0.08	-0.05 to 0.21	0.07	0.06			.21
2019-2020 cohort model					0.04	0.04	<.001
Constant	3.59	3.07-4.10	0.26	N/A			<.001
Sex ^c	0.17	0.07-0.27	0.05	0.18			<.001
Age	-0.00	-0.02 to 0.02	0.01	-0.02			.72
URM status	0.12	0.01-0.26	0.06	0.11			.04
2020-2021 cohort model					0.017	0.009	.12
Constant	3.65	3.18-4.12	0.24	N/A			<.001
Sex ^c	0.11	0.01-0.21	0.05	0.11			.04
Age	0.00	-0.02 to 0.02	0.01	0.02			.76
URM status	0.06	-0.06 to 0.17	0.06	0.06			.32
Total model					.035	.032	<.001
Constant	3.38	3.11-3.64	0.14	N/A			<.001
Sex ^c	0.15	0.09-0.20	0.03	0.15			<.001
Age	0.01	-0.002 to 0.02	0.01	0.05			.14
URM status	0.11	0.04-0.18	0.04	0.09			.003

^aMMI: multiple mini interview.

^bN/A: not applicable.

^cThe 2 “prefer not to answer” responses for sex were removed from analysis.

^dURM: underrepresented in medicine.

Table 5. Forward stepwise logistic regression predicting likelihood of acceptance to medical school.

Group, model	B	SE	Exp(B)	95% CI for Exp(B)
2018-2019 cohort				
Step 1				
Rank of readiness	1.083	.246	2.954	1.825-4.782
Constant	-3.162	.830	.042	N/A ^a
2019-2020 cohort				
Step 1				
Rank of readiness	.948	.212	2.580	1.702-3.912
Constant	-3.023	.739	.049	N/A
Step 2				
Negotiating the plan	.547	.215	1.728	1.133-2.637
Rank of readiness	.685	.234	1.983	1.253-3.139
Constant	-3.275	.752	.038	N/A
2020-2021 cohort				
Step 1				
Therapeutic relationship	.787	.201	2.196	1.480-3.258
Constant	-2.791	.878	.061	N/A
Step 2				
Opening the encounter	-.583	.272	.558	.328-.951
Therapeutic relationship	1.093	.251	2.984	1.823-4.884
Constant	-1.685	1.020	.185	N/A
Step 3				
Opening the encounter	-.792	.296	.453	.254-.808
Empathy	.552	.278	1.736	1.006-2.997
Therapeutic relationship	.834	.282	2.303	1.325-4.000
Constant	-1.940	1.035	.144	N/A
All cohorts combined				
Step 1				
Empathy	.688	.097	1.989	1.644-2.406
Constant	-2.265	.388	.104	N/A
Step 2				
Empathy	.414	.132	1.513	1.169-1.958
Rank of readiness	.552	.183	1.737	1.213-2.489
Constant	-3.087	.479	.046	N/A
Step 3				
Opening the encounter	-.334	.155	.716	.529-.971
Empathy	.519	.141	1.681	1.275-2.216
Rank of readiness	.690	.195	1.993	1.360-2.921
Constant	-2.573	.533	.076	N/A

^aN/A: not applicable.

For the 2018-2019 cohort, the model was statistically significant, ($\chi^2_1=21.33$; $P<.001$). The model explained 8.7% (Nagelkerke

R^2) of the variance in acceptance status and correctly classified 64.2% (206/321) of cases. Sensitivity was 89.9%, specificity was 22.1%, positive predictive value was 65.3%, and negative

predictive value was 57.4%. Rank of readiness was the only variable in the model.

For the 2019-2020 cohort, the final model was statistically significant ($\chi^2=28.59$; $P<.001$). The model explained 11% (Nagelkerke R^2) of the variance in acceptance status and correctly classified 64.3% (214/333) of cases. Sensitivity was 77.5%, specificity was 47.3%, positive predictive value was 65.3%, and negative predictive value was 62.2%. The final model (model 2) consisted of 2 statistically significant MMI variables: rank of readiness and negotiating the plan. Rank of readiness was associated with the greatest increase in the likelihood of being accepted to medical school.

For the 2020-2021 cohort, the final model was statistically significant ($\chi^2=24.97$; $P<.001$). The model explained 10.4% (Nagelkerke R^2) of the variance in acceptance status and correctly classified 68.2% (217/318) of cases. Sensitivity was 91.3%, specificity was 25.2%, positive predictive value was 69.4%, and negative predictive value was 60.9%. The final model (model 3) consisted of 3 statistically significant MMI variables: opening the encounter, empathy, and therapeutic relationship. Therapeutic relationship was associated with the greatest increase in the likelihood of being accepted to medical school.

The final model for all cohorts combined was statistically significant ($\chi^2=67.95$; $P<.001$). The model explained 9.2% (Nagelkerke R^2) of the variance in acceptance status and correctly classified 65.5% (637/972) of cases. Sensitivity was 87.9%, specificity was 30.6%, positive predictive value was 66.5%, and negative predictive value was 61.7%. The final model (model 3) consisted of 3 statistically significant MMI variables: empathy, rank of readiness, and opening the encounter. Rank of readiness was added in model 2 and was associated with the greatest increase in the likelihood of being accepted to medical school in the final model.

Discussion

Principal Findings

In this study, we demonstrated that SPs were able to develop a useful, credible, and relevant measure that can help the medical school admissions committee evaluate applicants beyond academic achievements. Although the MMI is only 1 portion of data used in a holistic review of medical school applicants, it can be useful to examine the impact and process of the MMI in the medical school admissions. Additionally, the created global readiness question that was incorporated into the MMI process contributed to selecting candidates for medical school.

Beginning with the 2018-2019 application cycle, simulation specialists integrated the revised rubric with the readiness question into training and conducting the MMI. This process was successfully adopted by the SPs involved in the MMI. As the SPs began using the new method of quantitatively scoring the applicant's readiness, they found this 1 question to be more efficient than providing qualitative feedback for each applicant. Since there are only about 3 minutes between applicant

encounters, the readiness question allows SPs to provide more concise feedback without being rushed.

Including stakeholders in the development of surveys and assessment instruments has been useful in other settings outside of simulation centers and has led to credible and relevant tools [47-50]. In this setting, working with SPs to create the rank of readiness question revealed what could be appropriately assessed through the lens of the SPs. The creation of the question through patient and public involvement yielded a more objective and standardized measure for scoring purposes. Additionally, stakeholder involvement contributed to a sense of value and co-ownership in the admissions process. Furthermore, by codeveloping this rubric item with the SPs, we built on their previous MMI skills, allowed item development to be iterative, allowed SPs to own the process, and accommodated their scoring needs. Incorporating these and other processes have been found to be associated with higher rates of positive research outcomes in cocreated projects [28,29].

The results of the workshop positively impact the simulation specialists providing a standardized scoring opportunity for all applicants. Training for standardization is critical in this role. Due to the collaborative nature of the workshop, the simulation specialists were able to emphasize the value of the SPs' voices and consistent use of the evaluation tool, which is consistent with findings in other settings [27,51]. Moreover, the involvement of the SPs in the development of the question facilitates coaching of new SPs by simulation specialists to use this question to validate or summarize the data from the other rubric sections.

Our results showed that the MMI overall and the readiness question specifically were able to predict medical school acceptance. This finding is consistent with previous findings showing that SPs are able to assess students' communication skills [52-55]. Furthermore, our findings are consistent with previous results showing that the SPs were unbiased in their assessment of the interviewees [10,56]. The observed interviewees' demographics (age, sex, and race) predicted only 3% of the SPs' MMI overall score. Although the findings were statistically significant, the predicted amount of change within the different demographic variables was relatively small. There is a possibility that bias was reduced because 2 SPs scored each interviewee collaboratively.

Although the MMI rank of readiness score was statistically significant for predicting an applicant's acceptance to medical school overall and for the 2018-2019 and 2019-2020 cohorts, this was not the case for the 2020-2021 cohort. One potential explanation for this lack of significance is that the entire cycle was conducted in a web-based format due to the COVID-19 pandemic. The rubric was not changed to reflect the web-based interview environment, which could potentially account for some of the differences. For example, students were automatically given points for elements that could not be performed in a web-based environment (eg, knocking on door prior to entering). Additionally, nonverbal behavior was challenging to assess, as illustrated by score differences between cohorts with in-person or web-based MMIs (Multimedia Appendix 2). Although medical schools have faced many

challenges in moving to a web-based interview format, it has been found that web-based interviews can be as reliable as traditional interviews in making sound decisions on applicants [57]. More research is needed to assess the impacts of the COVID-19 pandemic on medical school admissions and web-based SPs encounters.

Limitations

Despite the SPs' ability to create a measure, several other factors play a role in the admissions selection process beyond the MMI including, but not limited to, grade point average, Medical College Admission Test scores, recommendation letters, personal statements, and distance traveled (overcoming adversity or obstacles). These additional factors were not included in the models. Although the SP portion of the MMI excelled at predicting acceptance, specificity was low overall and for each cohort. Other elements of the application may have a greater impact on the applicants who were declined. Although the actual results of the statistical analysis might not be generalizable to other settings, lessons learned from the process for having patient and public involvement when creating a measurement instrument can benefit other institutions.

Future Implications

The standard rubric used for MMI may need to be revised to remove items that cannot be performed over the web (eg,

knocking on door, appropriate touch, and sustaining personal space) for admission years when web-based MMI is the standard. This potential, revised rubric could also be used to assess learners during telehealth encounters, and the new rubric would need to be evaluated. Additionally, the MMI rubric without the final readiness question is currently being used as the communication rubric for assessing learners during standard SP encounters and Objective Structured Clinical Examinations. The global question is specifically intended for SPs to determine applicant readiness for medical school, so it should not be used for students who have matriculated. However, it could be beneficial to replicate the workshop with SPs to identify a global question for the communication rubric. Finally, an additional investigation could be conducted to see how matriculating students' MMI scores compare with the future communication scores throughout their time in medical school.

Conclusion

The collaborative nature of this project between stakeholders, including nonacademics and researchers, was vital for the success of this project. This study shows that SPs bring a critical perspective that can improve the admissions process of evaluating medical school applicants through the MMI process. They also can further incorporate themselves as team members by cocreating an effective global question to improve the evaluation of the applicants.

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For this paper, we included representation from all stakeholders in the authorship process, including students (current student, matriculant, and applicant), simulation specialists, standardized patients, researchers, admissions committee members, and admissions department staff.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Workshop presentation for standardized patients to create a global assessment scale for the multiple mini interview. [[PDF File \(Adobe PDF File\), 4328 KB - jopm_v14i1e38209_app1.pdf](#)]

Multimedia Appendix 2

Multiple mini interview rubric, preworkshop and postworkshop. [[PDF File \(Adobe PDF File\), 365 KB - jopm_v14i1e38209_app2.pdf](#)]

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Abbreviations

- MMI:** multiple mini interview
SP: standardized patient
URM: underrepresented in medicine

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Original Paper

Implementing the Co-Immune Open Innovation Program to Address Vaccination Hesitancy and Access to Vaccines: Retrospective Study

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Abstract

Background: The rise of major complex public health problems, such as vaccination hesitancy and access to vaccination, requires innovative, open, and transdisciplinary approaches. Yet, institutional silos and lack of participation on the part of nonacademic citizens in the design of solutions hamper efforts to meet these challenges. Against this background, new solutions have been explored, with participatory research, citizen science, hackathons, and challenge-based approaches being applied in the context of public health.

Objective: Our aim was to develop a program for creating citizen science and open innovation projects that address the contemporary challenges of vaccination in France and around the globe.

Methods: We designed and implemented Co-Immune, a program created to tackle the question of vaccination hesitancy and access to vaccination through an online and offline challenge-based open innovation approach. The program was run on the open science platform Just One Giant Lab.

Results: Over a 6-month period, the Co-Immune program gathered 234 participants of diverse backgrounds and 13 partners from the public and private sectors. The program comprised 10 events to facilitate the creation of 20 new projects, as well as the continuation of two existing projects, to address the issues of vaccination hesitancy and access, ranging from app development and data mining to analysis and game design. In an open framework, the projects made their data, code, and solutions publicly available.

Conclusions: Co-Immune highlights how open innovation approaches and online platforms can help to gather and coordinate noninstitutional communities in a rapid, distributed, and global way toward solving public health issues. Such initiatives can lead to the production and transfer of knowledge, creating novel solutions in the public health sector. The example of Co-Immune contributes to paving the way for organizations and individuals to collaboratively tackle future global challenges.

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KEYWORDS

open science; open innovation; programmatic research; collective intelligence; web based; immunization; vaccination access; vaccine hesitancy; innovation; vaccine; public health; access; framework; participatory; design; implementation

Introduction

Background

As the world faces a rise in the number of complex challenges that threaten the resilience of our economic, environmental, social, and health systems, we observe a shift toward more collaboration and openness in the way science and innovation is performed [1-3], bringing governments, civil society, and the private sector closer. Examples of this include the efforts made to accelerate society's progress toward the Sustainable Development Goals (SDGs) [4] and the fight against pandemics, such as COVID-19 [5]. Yet, access to vaccines and vaccination hesitancy remain as some of the complex challenges to be addressed in order to achieve universal health coverage [6].

Immunization is one of the most cost-effective interventions to protect oneself and others from infectious diseases [7] and saves between 2 million and 3 million lives per year [8].

Yet, the annual death toll for vaccine-preventable diseases stands at 1.5 million, and large gaps in coverage persist, not only between countries but also within their territories [7]. In particular, the World Health Organization (WHO) listed vaccine hesitancy among the top 10 global health threats for 2019 [9]. Continuing global efforts to leave no one behind may be a long-standing challenge [10] when new information technologies and social media platforms are both part of the problem [11] and the solution. More recently, the COVID-19 pandemic demonstrated the repertoire of logistical and administrative challenges to the deployment and administration of vaccines, especially in low-resource settings [12].

In response, the WHO Global Vaccine Action Plan 2011-2020 [7] committed 140 countries and 290 organizations to promoting and prioritizing greater collaboration between governments, nongovernmental organizations, the private sector, and all citizens to address outbreaks of vaccine-preventable diseases. Additionally, a number of new digital and open innovation initiatives have been launched: the WHO has developed the Vaccine Safety Net [13], a network of websites about vaccination; health authorities in Canada have developed a school-based quiz to educate children about immunology and vaccines [14]; Finland is testing a computer game to communicate the benefits of human papillomavirus vaccination [15]; a project in India uses digital necklaces to record children's immunization history [16]; and the global Vaccination Acceptance Research Network has been established [16].

Global health guidelines showcase the positive outcomes of social participation for universal health coverage [17], which include more meaningful dialogue, more sustainable solutions, and more trust from citizens in health system institutions or in the decisions that are made. Indeed, there is room for more initiatives that allow people to genuinely co-design solutions in a multidisciplinary manner during and following pandemics [18]. Hence, the number and sustainability of these types of

initiatives could be amplified by fostering increased collaboration with nonacademic citizens in the creation and development of solutions in an open innovation framework [19]. This is the gap that Just One Giant Lab (JOGL) is proposing to fill with the Co-Immune program.

Citizen science is an emerging and highly diverse practice that can be broadly defined as the general public being involved in the process of doing research [20]. Research has demonstrated that intensity and diversity of collaboration positively affect the quality [21] and productivity [22] of research, while positively impacting the knowledge integration from participants [23]. Likewise, participant transdisciplinarity [24] seems critical to generating innovative outcomes [25] and dealing with complex real-world problems [26]. Such mechanisms are often at play in the field of citizen science, promising to transform the knowledge generation landscape by tapping into networks of nonacademic citizens [26,27] in a new social contract for this kind of research [28]. Citizen science has the potential to expand the number of individuals contributing knowledge and ideas, transform how hypotheses are generated, and transform how data sets are analyzed. Such approaches have already been applied to investigate individual diseases through patient-led research [29,30] and public health challenges, such as the epidemiology of cancer [31-33].

Other approaches to create and develop knowledge and solutions to complex challenges are slowly entering the mainstream. In particular, hackathons, challenge-based approaches, and the participation of citizens in science have been flourishing over the last two decades [34], especially within the natural sciences [35] and, more recently, within medical sciences, public health, and population-health research [36,37].

Hackathons are short, intensive, and collaborative events that are designed to prototype solutions addressing a specific problem. They originated in the early 2000s in digital and tech fields and have been adapted to address more complex challenges in global health [38-40]. Such initiatives are not without pitfalls: they suffer, by design, from the lack of paths to sustainability for the projects they launch [41]. In response to such criticisms, there are increasing efforts, such as the "Make the Breast Pump not Suck" hackathon and "Trans*H4CK," to improve hackathon methodology by working directly with affected communities [41]. Several initiatives, such as a Massachusetts Institute of Technology collaborative design studio, provide insights into hackathon methods [42] to facilitate better hackathons [43,44]. More recently, multiple entities have engaged in organizing hackathons to address the COVID-19 crisis [45,46].

Challenge-based approaches, which provide frameworks for learning while solving real-world issues, have also been on the rise in global health and have proven to be efficient for generating innovative solutions and for incentivizing mass community engagement [45]. For example, the potential of participative models to address complex questions, along with

the power of contests to offer a structure that catalyzes this work, has been exhibited by the Epidemium initiative on cancer epidemiology [46].

Despite the numerous tools and technologies created to facilitate collaboration in citizen science projects, challenges remain. These include the issues of the complementarity, coherence, and diffusion of these initiatives [34] to efficiently address international policies and local needs, as the local adoption of hackathon solutions often remains low [47].

Therefore, the promotion of transdisciplinarity and citizen science in an open innovation framework, coupled with methods such as hackathons, and a challenge-based approach represent an opportunity to address current complex challenges of vaccination that would overcome the limits of either solution alone. In this paper, we describe the design, implementation, and outputs of Co-Immune, a collaborative open innovation program that was run in 2019 to address vaccination hesitancy and access to vaccination.

Objectives

Co-Immune's aim was to develop an environment that favors the creation and development of citizen science and open innovation projects addressing the contemporary challenges of vaccination in France and around the globe. This program had four specific objectives: (1) to foster a collaborative, open, and transdisciplinary dynamic; (2) to promote the emergence of accessible knowledge and innovative solutions; (3) to support participants in the elaboration and development of their project; and (4) to disseminate the outputs and results in an open science framework. In this study, we describe the methodology of Co-Immune and its implementation, and we present its key outcomes.

Methods

Design

The overall program duration was 10 months (March 2019 to January 2020), divided into 6 months of preparation and 4 months of rollout of activities that included offline and online events, support for the development of citizen science projects, and assessment and awards for projects participating in the challenge-based competition. The main outputs of the program were projects, categorized as leading to (1) knowledge production, if they performed data analysis or generated new knowledge, whether it was context specific, generic [48], or knowledge transfer [49]; or (2) solutions, such as hardware, software, and interventions.

Co-Immune was coordinated online through the JOGL platform [50] and supported by 13 partners from the public and private sector (Table S1 in [Multimedia Appendix 1](#)). The challenge-based nature of the program was designed to be an

incentive for teams and participants to continue developing their projects after hackathon events or to create their project on JOGL at any other time.

The governance of Co-Immune was designed to provide freedom for projects to develop innovative solutions while ensuring their compliance with local and international regulations and consideration of ethical and scientific integrity. To this end, we constituted the independent Committee for Ethics, Science and Impact (CESI), which issued an opinion on the rules of participation in the program and validated the strategic orientation of the program. Public health priorities were identified based on a literature review and divided between two main challenges to streamline participants' work: vaccination coverage and vaccination hesitancy. They were then validated by the CESI. In addition, through a series of semistructured interviews, experts at the 7th Fondation Merieux Vaccine Acceptance conference [51] identified eight specific issues to address and potential room for solutions. The CESI also participated in the co-elaboration of the assessment grid, which was used as a base to grant nonmonetary prizes to projects in December 2019.

To be eligible for a prize, a project was required to have created a comprehensive description of their initiative on the JOGL platform and a video pitch. This material was provided to experts in charge of the assessment.

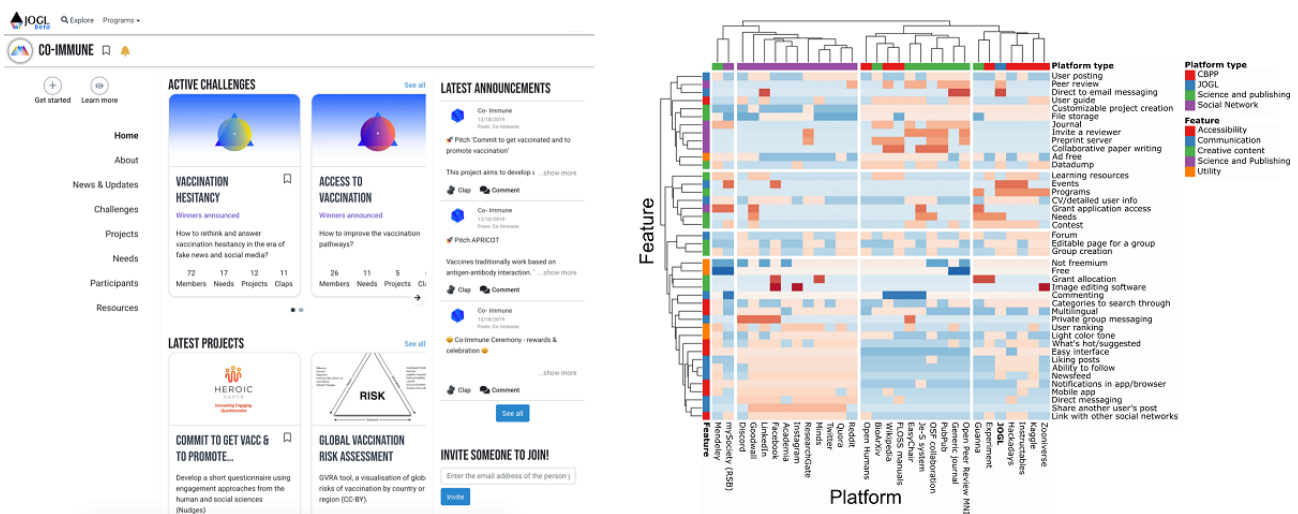
Participant Recruitment

Participants were recruited through our network of partners from around the globe and social media communication. Participation was open to everyone above the age of 18 years, if they had agreed to follow the participation rules validated by the CESI. Participants could take the role of "project leader" or "contributor."

JOGL Platform

Co-Immune participants used the JOGL platform to document their projects and recruit collaborators throughout the course of the program. JOGL is a decentralized mobilization platform designed for use in collaborative research and innovation ([Figure 1](#)). Within the JOGL platform, users can create a profile and declare their skills. Once registered, they can create or join projects, follow the activity of other members, post on their project feed, and comment on other posts. They can also highlight needs for a project they are part of, specifying skills that can help to solve project problems. We compared the JOGL features to those of other online platforms for citizen science, social networking, and science and publishing through a cluster analysis (see [Figure 1](#) as well as the supplementary method and [Figure S1](#) in [Multimedia Appendix 1](#)), indicating that the platform is functionally similar to other platforms in the space and is suitable to hold a citizen science program such as Co-Immune.

Figure 1. Overview of the Just One Giant Lab (JOGL) platform. The image on the left is a screenshot of the JOGL platform. The right-hand image is a heatmap of feature presence across popular online tools. For each platform (columns), we numerically encoded the presence (1) or absence (0) of each feature (rows). Then, for each element, we computed a Z score by standardizing values across platforms, represented here by the color spectrum: blue (low) to red (high). CBPP: citizen-based peer production network (ie, citizen science platform); CV: curriculum vitae; Je-S: Joint Electronic Submissions; MNI: Montreal Neurological Institute; OSF: Open Science Framework; RSB: Royal Society of Biology.

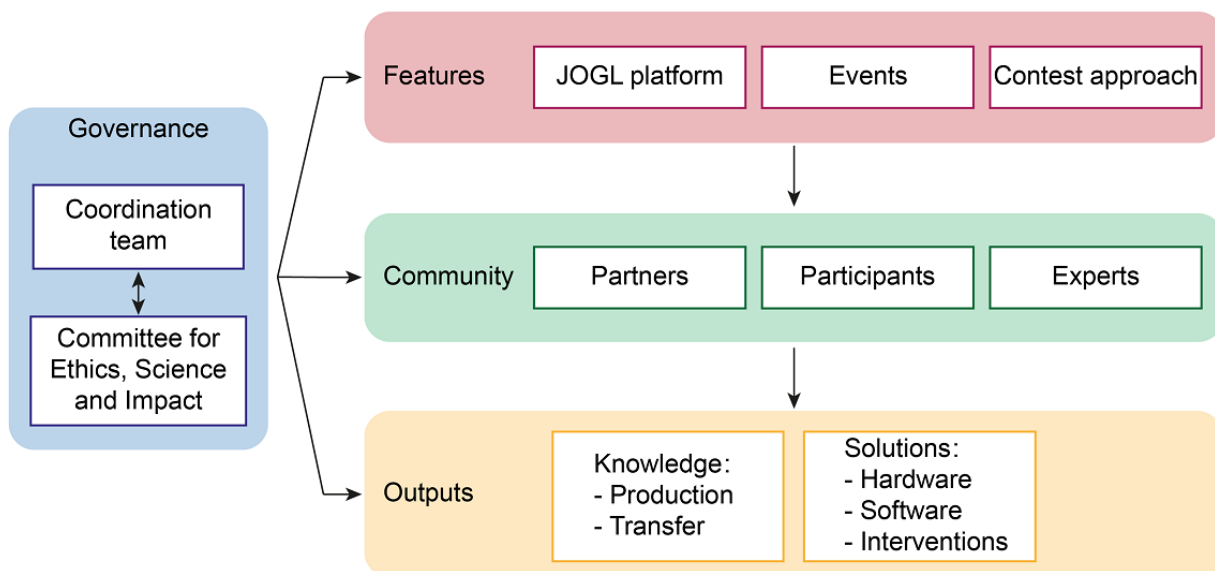


Implementation

The Co-Immune program was realized through an interrelated and interacting set of technological and social features (Figure 2). Our coordination team implemented the larger program (ie, events, online platform, and contest approach) and helped to recruit a community of partners and participants who interacted

with each other and were supported in their efforts through the high-level design features. With support from the governance structure of the Co-Immune program, the individual projects managed to provide outputs that included knowledge production and transfer as well as solutions, such as hardware, software, and interventions.

Figure 2. Workflow of the Co-Immune program design. JOGL: Just One Giant Lab.



Building an Open Community

To build the community, we contacted organizations involved in a wide range of domains before the launch of the program, thereby creating a first pool of contributing professionals and students. We also recruited participants via the organization of events, typically in the evening, aimed at creating projects, fostering collaboration among participants to address project

needs, and providing mentorship. To facilitate the coordination of the community, all participants were required to use the JOGL platform to describe their projects, form teams, list their needs, and initiate collaboration.

In order to create a supportive and collaborative environment for the participants, we reached out to various organizations to establish partnerships. Our intention was two-fold: (1) to

facilitate the participation of the organizations’ students and employees as participants or mentors by involving their institution and (2) to enhance the sustainability of projects after the course of the program by connecting them with potential partners at the early stage of their development.

The 13 partners operated in the health, technology, and social sectors, and included research, innovation, and education organizations, as well as professional networks, incubators, and communication specialists (Figure 3). The number of partners grew over the life span of the initiative and were often suggested by existing partners or through connections made during events.

We organized 10 offline and online events between October and December 2019 (Table 1). Participants for events were recruited through social media and mailing lists leveraging our network of partners. Among the four on-site events that were organized, two were hackathons aimed at motivating participants to join the program, while the other two were aimed at fostering collaboration around the most advanced projects. Their median duration was 2.25 (IQR 2) hours.

The facilitation of the hackathon-style events relied on the use of participatory and collective intelligence design and problem-solving techniques [52]. In particular, participants were encouraged to form multidisciplinary teams including both professionals and students.

Three partners in Paris—Epitech, the Wild Code School, and the Center for Research and Interdisciplinarity (CRI)—co-organized and hosted events for their students, respectively, in their engineering, coding, and life science and education schools. Other partners—Kap Code, Excelya, and CorrelAid—mobilized their teams to act as mentors during these events. A total of 14 mentors attended events, and five came to more than one event.

In addition, we organized four 1-hour online events. The first was an opportunity to share information about Co-Immune with people around the globe. Another event discussed best practices to document open science projects. Finally, two events focused on the resolution of needs of single projects (Table 1 [53,54]).

Figure 3. Treemap representing the domains of action of the 13 Co-Immune partners.

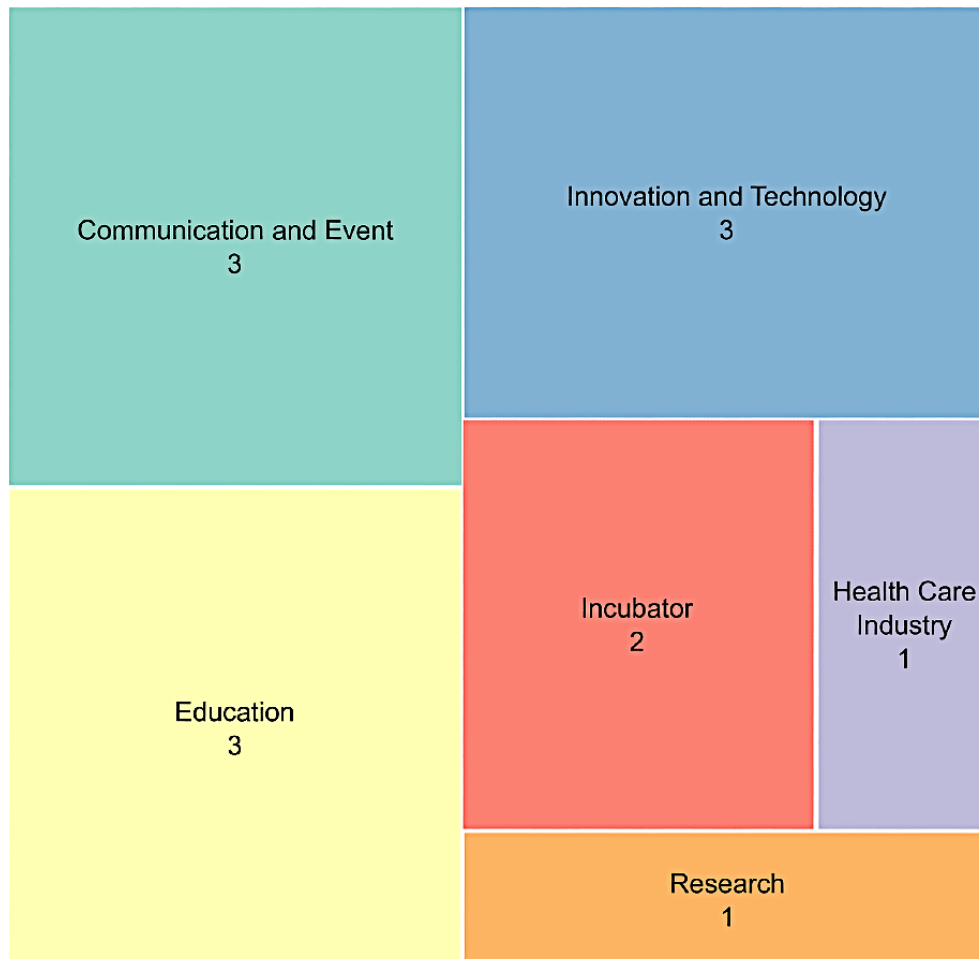


Table 1. Co-Immune events.

Name	Mode; type; location	Duration (hours), n	Objective	Design; supporting partners (if applicable)	Participants, n
Launch	Offline; ceremony; CRI ^a , Paris	3	Gather the initial community	Presentation of the program design, features, timeline, and partners, as well as networking	60
OpenJOGL ^b ; Co-Immune	Online	1	Q&A ^c session on the program	Presentation of Co-Immune and questions and answers	3
Sprint; open data	Offline; hackathon; CRI, Paris	2.5	Build community, create projects, and create data repositories	Statement of the problem (videos of experts), team formation and effort, mentoring, and publication of results on the JOGL platform; supported by CRI and CorrelAid	25
OpenJOGL; Vaccination Awareness Escape Game [54]	Online	1	Foster collaboration around single projects	Pitch of the project and its needs, feedback from experts, and questions and answers	7
Sprint; project creation	Offline; hackathon; CRI, Paris	4	Build community and create multidisciplinary projects	Statement of the problem (videos of experts), ice breaker, multidisciplinary team formation and effort, mentoring, presentation of results, vote for the most promising projects, publication of results on the JOGL platform, and networking; supported by CRI, Epitech, Wild Code School, CorrelAid, and Excelya	22
Sprint; open data	Offline; hackathon; Wild Code School, Paris	3	Accelerate the development of projects related to data science	Selection of a project by participants among the two choices available, team formation and effort, mentoring, presentation of results, publication on the JOGL platform, and networking; supported by Wild Code School, CorrelAid, and Excelya	15
Sprint; open data	Offline; hackathon; Epitech, Paris	3	Build the community, create projects, and accelerate the development of one project using Twitter data	Statement of the problem, selection of a project by participants among the four choices available (including one already existing project), team formation and effort, mentoring, presentation of results, vote for the most promising project, publication of results on the JOGL platform, and networking; supported by Epitech, Kap Code, Excelya, and CorrelAid	35
OpenJOGL; HERA ^d : A Health Platform for Refugees [53]	Online	1	Foster collaboration around single projects	Pitch of the project and its needs, feedback from experts, and questions and answers	7
OpenJOGL; better documentation for better collaboration	Online	1	Help teams document their projects in the most open and reproducible way	Expert presentation on best practices for documenting open science projects, presentation of Co-Immune expectations for documentation, and questions and answers	13
Closing ceremony	Offline; ceremony; CRI, Paris	2	Close the Co-Immune program	Presentation of the main outputs of the program and awards for the best projects	70

^aCRI: Center for Research and Interdisciplinarity.

^bJOGL: Just One Giant Lab.

^cQ&A: question and answer.

^dHERA: Health Recording App.

Co-Immune Experts: CESI Members, Mentors, and Interviewees

Individuals who were considered “experts” included all the CESI members as well as experienced professionals of a certain field who attended events and provided technical guidance to teams as “mentors.”

The CESI members were sought to represent the diversity of stakeholders involved in advancing access to vaccines and reducing vaccine hesitancy. By choosing interviewees who were

researchers specializing in the challenges of access to vaccines and vaccination hesitancy, we aimed at benefiting from their expert understanding of the issues and of the priorities to be addressed to streamline the work of participants around particular problems. Finally, we grew the pool of mentors over the span of the program to best match their expertise with the needs of the projects in an agile manner.

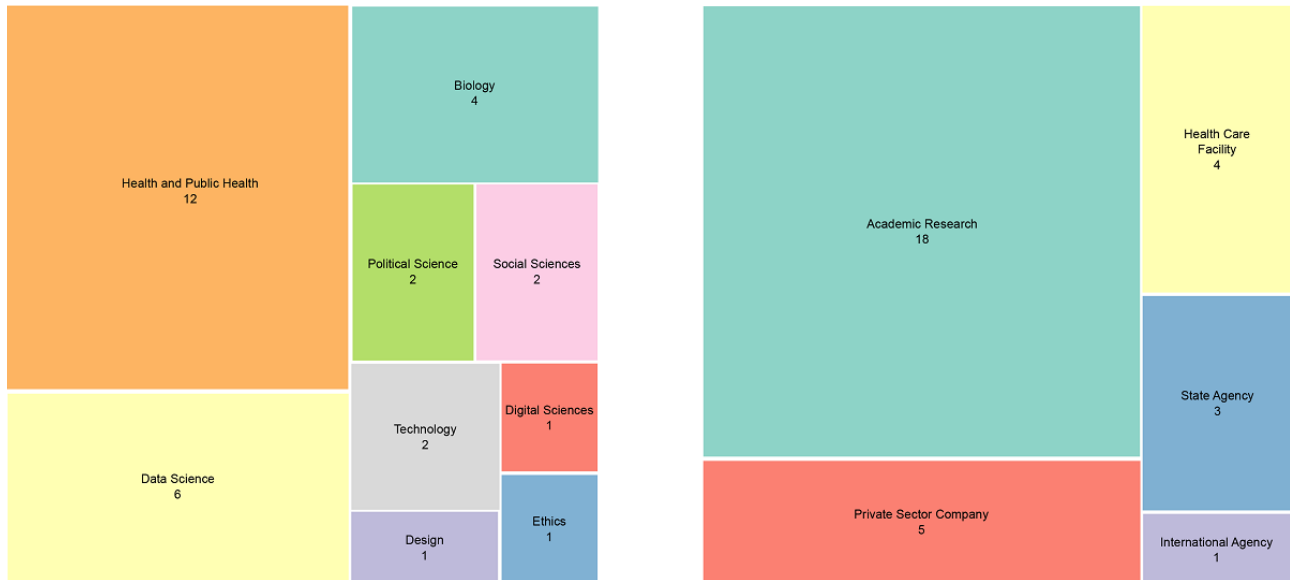
Overall, the mentors’ domains of expertise ranged from biology to social sciences, design, technology, and data science (Figure

4). One-third of them were working as health or public health professionals.

The CESI consisted of eight volunteer members and included virologists, pharmacists, health economists, experts in the digital

sciences and ethics fields, and biologists; members were working at international, national, and local levels of the health system. All of them worked for public or nonprofit organizations. Interviewees were mostly researchers in social sciences and medical practitioners.

Figure 4. Treemap of the 31 Co-Immune experts: domains of expertise (left) and affiliations (right).



Co-Immune Project Assessment

The assessment of projects by experts was designed to be an opportunity for learning and growth. To be assessed, teams were asked to provide a video pitch summarizing their project and detailed documentation on their project page on the JOGL platform, including links to their open access data and code. Project assessment was performed through a grid that was codeveloped by JOGL and the CESI. In addition to grades, teams received detailed feedback on their projects.

The assessment grid was based on a literature review of project evaluation standards and consisted of 10 questions graded from 0 to 5 (Multimedia Appendix 2). Three areas were assessed: the approach, the implementation strategy, and the impact. First, the assessment of the approach included the following: (1) clarity and relevance of the problem and alignment with the program scope, (2) fit between the approach and methodology and the problem statement, and (3) innovation potential (ie, the project introduces groundbreaking objectives, novel concepts, or approaches). Second, the implementation strategy was assessed using following the criteria: (1) state of progress toward set goal (ie, state of advancement), (2) clarity and relevance of the timeline and needs for the future (ie, major tasks and milestones), and (3) project actively engages and aligns with all relevant stakeholders. Finally, the assessment of the impact covered the following: (1) clarity and relevance of the criteria used to measure impact, (2) the extent to which the project considers its ecosystem (ie, ecological, environmental, ethical, and social considerations), (3) sustainability and scalability of the project in the long term, and (4) open and reproducible dissemination strategy. For each of these three categories, JOGL awarded a prize to the project with the best score based on the grades given by reviewers. Additionally, a grand prize was given

to the project with the overall highest score. JOGL provided visibility, while two partners also provided an award to a project of their choosing.

JOGL Platform Data Collection and Analysis

Participants added their professional background, skills, and employment status to the JOGL platform. These data were used to evaluate the composition of the community. All users who joined JOGL during the span of the program were considered to be participants of Co-Immune, as it was the only ongoing program, and all outreach activities were related to it.

To better understand how skills were related across participants, we used a network approach to assess similarity between skills and to get further insights about the global diversity of the community. In this network approach, each declared skill was a node and the skills were considered linked if they co-occurred in a participant. Links were then weighted by the number of participants within which they co-occurred. Gephi 0.9.2 was used to represent the network shown in the skill map of the Co-Immune community, and the modularity algorithm was used with default parameters to compute communities representing the sets of skills that tend to co-occur more together than with other skills. Since these skills are linked through the participants who share them, they can be understood as "participant types" constitutive of the Co-Immune community.

We provide the data related to this study on Zenodo [55]. These data include (1) the link, description, and assessment scores of projects; (2) the profiles of platform users; (3) the description of events; (4) the profiles of experts; and (5) the list and types of partners.

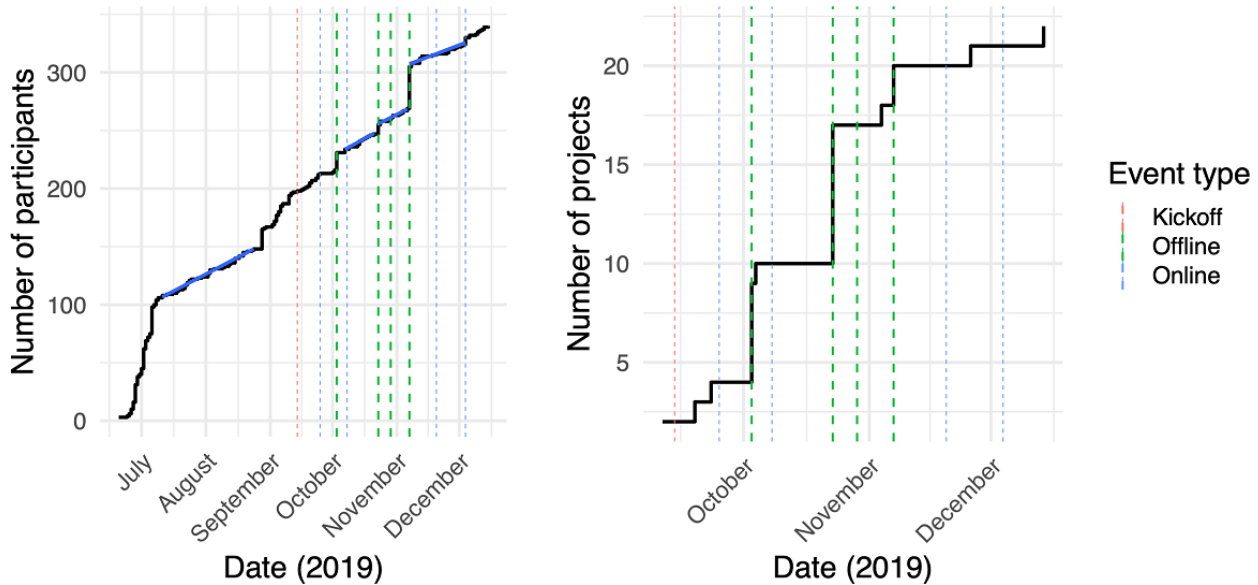
Results

Community Growth Through Events

During the program, 234 participants signed up to the platform (Figure 5). The participant growth was mostly linear over the life span of the program (July 10 to December 18, 2019), suggestive of the potential for continued growth if the program had continued. The growth rate outside of events, at around one

per day (between 0.86 and 0.98 users/day), was consistent with the prekickoff growth rate (0.94 users/day). This highlights the importance of events (dashed lines in Figure 5) for driving participant enrollment, with the four offline events accounting for 45% of the growth. In total, offline events were responsible for the generation of 82% (18/22) of the projects. The rest consisted of 4 out of 22 (18%) projects created on the platform outside of events and 2 already-existing projects prior to the program.

Figure 5. Growth of the number of participants (left) and number of projects (right) over the life span of the program. Dashed bars show when events for community facilitation were held (green: offline events; blue: online events; red: kickoff meeting). Blue lines give a linear fit during the corresponding periods, showing stable growth pre- and postkickoff.



Participant Skills and Backgrounds: A Transdisciplinary Community

Out of the 234 participants, 187 (79.9%) declared their job category. The community was composed of a mix of students (67/187, 35.8%) and workers (94/187, 50.3%), most of whom worked full time (81/94, 86%; Figure 6). Other categories included “between jobs” (n=11), “nonprofit” (n=12), and “for profit” (n=3). Out of the 75 participants who declared their country in their JOGL profile, 57% (n=43) were based in France, with the rest coming from other regions, including the rest of Europe, the Americas, Africa, and Asia.

The 234 participants specified a total of 492 unique skills (median 3 [IQR 4.5] skills per participant). We observed a high representation of data science and coding alongside biology, which, altogether, related to the technical skills emphasized during the program (Figure 6). The skill network shows that the community spanned a vast interdisciplinary landscape, from open science to open data and coding, and from project management to biology. The network exhibited the largest connected component of 416 interconnected skills (84.6% of all skills; Figure 7). The modularity maximization (see the Methods section) resulted in the identification of 12 modules corresponding to “participant types” constitutive of the Co-Immune community.

Figure 6. An overview over the Co-Immune community: participant categories (left) and the 20 most represented skills (right) in the Co-Immune community.

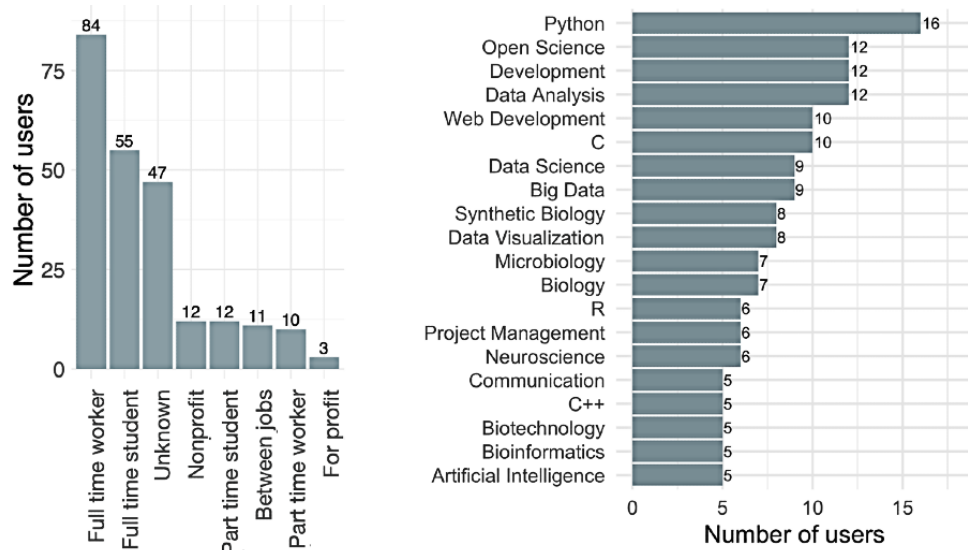
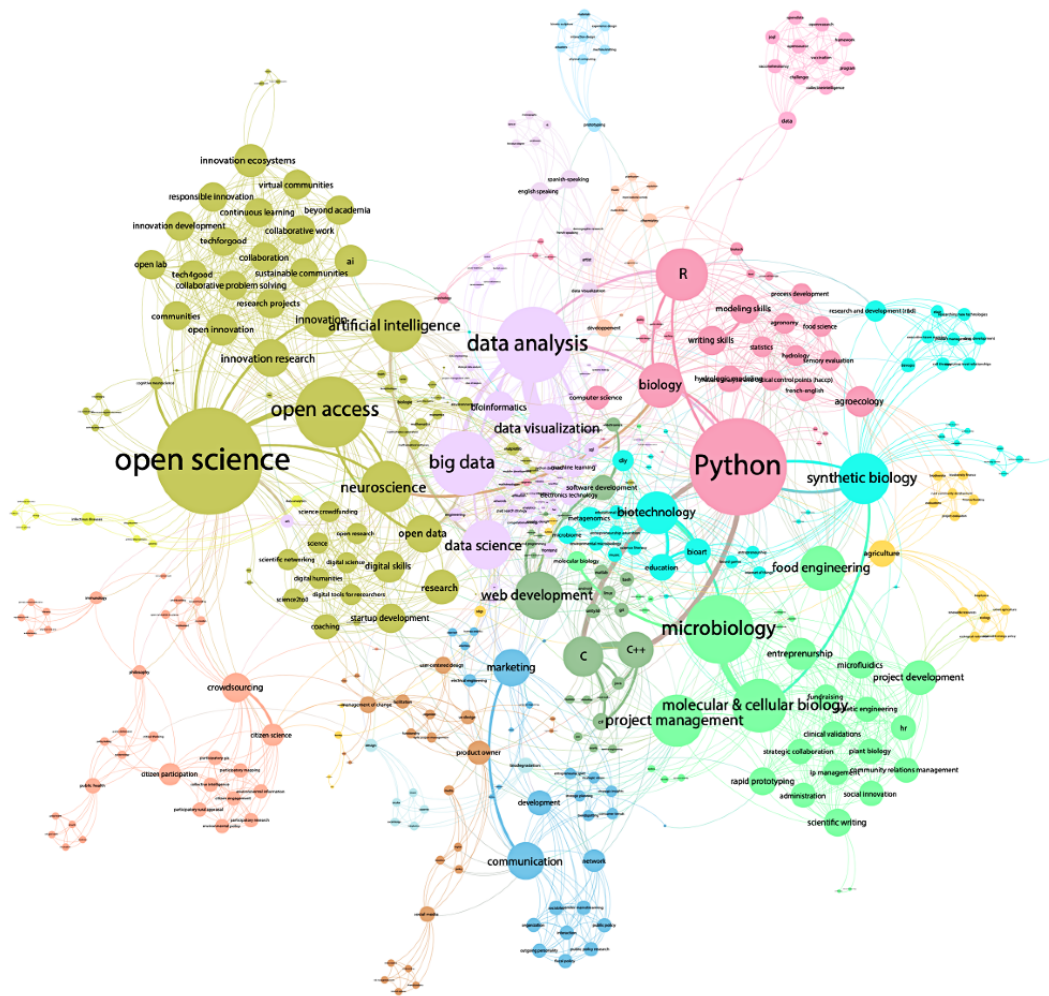


Figure 7. Skill map of the Co-Immune community. Skills are linked if they appear in the profile of the same participant. Link weight indicates the number of participants sharing the skills. Node size indicates weighted degree.



Co-Immune Project Description

A total of 22 projects were created by 20 project leads, with teams of up to 11 members (Table 2 [52,53,56-75]). Among these, 15 (68%) projects proposed to develop software covering

web technologies, mobile apps, algorithms, data lakes, data modeling and analysis, and visualization tools. The other 7 (32%) projects included hardware development and interventions involving biotechnologies, game design, behavioral

sciences, education, and communication. Overall, one-third of the projects focused on knowledge transfer.

Among the 15 projects relying on software technology, 11 (73%) aimed at contributing to the production of knowledge by facilitating the analysis of publicly available data; they did this via the use of parsing tools and the creation of repositories (n=3), the analysis of open data (n=3), the development of machine learning tools to extract and analyze Twitter data related to vaccination hesitancy (n=2), and the production of data visualizations (n=3). In particular, more than 40 data sets were identified and collected by 4 projects that were created during the data-centered events. In addition, a database of 2464 tweets, in French, posted over a period of 7 years was made available by a partner, and another data set of 89,979 tweets was gathered by the project Qualitative Analysis of Tweets on Vaccination [56].

Out of the 15 projects above, 4 (27%) used software for knowledge transfer; for instance, the HERA (Health Recording App) project [52] provided educational content and health data

storage through its mobile app to improve the monitoring of vaccination and perinatal health among Syrian refugees in Turkey. The Pass It On project [60] focused on role-playing video games directed at health professionals as another method of knowledge transfer. The Neutralizing Information About Vaccines project [70] implemented an algorithm for parsing web pages, helping citizens identify trustworthy content related to vaccines.

A total of 5 projects out of 22 (23%) focused on different interventions (Table 2), including raising awareness about vaccination through an escape game (ie, Vaccination Awareness Escape Game [54]) and communication campaigns on social media (ie, Go Viral! [71]). The HEROIC Santé project [57] developed and tested a short questionnaire using engagement approaches from the social sciences to engage health care professionals and users around the question of flu vaccination. Finally, one team proposed applying synthetic biology methods to tuberculosis vaccines (ie, Project APRICOT [Antigen Presentation Using Crispr for TB] [58]).

Table 2. Co-Immune project descriptions.

Project name	Project status	Solution category	Summary description
HERA ^a : A Health Platform for Refugees [53]	<ul style="list-style-type: none"> Assessed^b Awarded Grand prize Best approach prize Best impact strategy prize 	<ul style="list-style-type: none"> Software Knowledge transfer 	A mobile health app designed for improving the monitoring of vaccination and perinatal health of Syrian refugees in Turkey; it provides recall of vaccines, storage of health data, health promotion (educational content), and financial incentives for immunization
Qualitative Analysis of Tweets on Vaccination [56]	<ul style="list-style-type: none"> Assessed Awarded Partner prize 	<ul style="list-style-type: none"> Software Knowledge production 	A web-based platform providing real-time visualization and analysis of tweets related to vaccination and vaccination hesitancy; data analysis included sentiment analysis and network analysis; an area of development was the development of predictive models of epidemic occurrence based on Twitter data
Commit to Get Vacc & to Promote Vaccination – HEROIC Santé [57]	<ul style="list-style-type: none"> Assessed Awarded Best implementation strategy prize 	<ul style="list-style-type: none"> Intervention Knowledge transfer 	A short questionnaire (7 minutes) using engagement approaches from the human and social sciences, such as “the importance of the source,” “voluntary consent,” or “fear and danger management,” to engage health care professionals and users, not only to be vaccinated against the flu, but also to promote flu vaccination
Project APRICOT ^c [58]	<ul style="list-style-type: none"> Assessed Awarded Partner prize 	<ul style="list-style-type: none"> Hardware 	Development of a synthetic biology–based methodology that addresses the evasion mechanisms adopted by the mycobacterium tuberculosis and induces the acceleration of lysosomal biogenesis to improve antigen presentation
Vaccination Awareness Escape Game [54]	<ul style="list-style-type: none"> Assessed Not awarded 	<ul style="list-style-type: none"> Intervention Knowledge transfer 	An escape game to raise vaccination awareness among the general population
Harmonize Vaccination [59]	<ul style="list-style-type: none"> Assessed Not awarded 	<ul style="list-style-type: none"> Software Knowledge production 	A tool for parsing various formats of vaccination coverage data sets and for visualizing them on a common platform
Pass It On: A Game About Vaccine Hesitancy [60]	<ul style="list-style-type: none"> Assessed Not awarded 	<ul style="list-style-type: none"> Software Knowledge transfer 	A role-play video game aiming to improve the capacity of health professionals to respond to their patients’ hesitation to be vaccinated
Global Vaccination Risk Assessment [61]	<ul style="list-style-type: none"> Assessed Not awarded 	<ul style="list-style-type: none"> Software Knowledge production 	A tool to create an overview of risk factors of “not getting vaccinated,” by country, while looking at the more comprehensive picture; the methodology of this project is based on fuzzy logic, multi-criterion analysis, and the risk triangle
Immuno [62]	<ul style="list-style-type: none"> Not assessed^d 	<ul style="list-style-type: none"> Hardware Knowledge transfer 	A board game providing access to the general public’s understanding of medical sciences related to immunization
Vaccine DataDump [63]	<ul style="list-style-type: none"> Not assessed 	<ul style="list-style-type: none"> Software Knowledge production 	A vaccination-related data repository and analysis tool for quick analysis of vaccine-related issues
Measuring Vaccination Hesitancy From Social Media [64]	<ul style="list-style-type: none"> Not assessed 	<ul style="list-style-type: none"> Software Knowledge production 	Data analysis of social media (ie, Twitter) to examine whether negative sentiment related to vaccination precedes declaration of symptoms and to study the relationship between vaccination hesitancy and epidemiological outbreaks
Mortality According to Access to Vaccines [65]	<ul style="list-style-type: none"> Not assessed 	<ul style="list-style-type: none"> Software Knowledge production 	Data analysis exploring the link between immunization coverage, mortality rate, and distance from health centers
The Health System Matrices [66]	<ul style="list-style-type: none"> Not assessed 	<ul style="list-style-type: none"> Software Knowledge production 	Exploratory analysis of the various parameters influencing vaccination coverage over time
Meta Immune – Data Exploration of Existing DB [67]	<ul style="list-style-type: none"> Not assessed 	<ul style="list-style-type: none"> Software Knowledge production 	A data lake on immunization data

Project name	Project status	Solution category	Summary description
Biloba ^e [68]	• Not assessed	• Intervention	An intervention incentivizing people to increase vaccine uptake through vouchers, supporting the existing mobile app Biloba
Wakuchin Senshi [69]	• Not assessed	• Intervention • Knowledge transfer	An interactive role-play board game to increase awareness about vaccination among the general population
Neutralizing Information About Vaccines [70]	• Not assessed	• Software • Knowledge transfer	An algorithm for parsing web pages, identifying misinformation, and identifying trustworthy content to help users in their health decisions related to vaccines; this also aims to be used by search engines in their recommender systems
Go Viral! [71]	• Not assessed	• Intervention • Knowledge transfer	A communication campaign on social media using gamification methods to illustrate contagion among users and, thereby, increase awareness of the importance of vaccines
Make Vaccines Affordable [72]	• Not assessed	• Software • Knowledge transfer	A web-based portal with data related to population demand for care in order to negotiate prices of vaccines with suppliers
Identify Topics of Discussion in Vaccination Posts [73]	• Not assessed	• Software • Knowledge production	Analysis of discussion in vaccination-related posts on Twitter and their evolution over time
Detect Vaccine Administration in Social Media Patient Data [74]	• Not assessed	• Software • Knowledge production	A classifier able to detect vaccine administration in tweets related to vaccination
Detect Vaccine Hesitancy in Social Media Patient Data [75]	• Not assessed	• Software • Knowledge production	A classifier able to detect vaccine hesitancy in tweets related to vaccination

^aHERA: Health Recording App.

^bThese were projects that were assessed by experts at the end of the program. To be assessed by a pool of experts, the project team needed to provide detailed documentation of their project, provide a short video pitch, and deposit their data and code on the Just One Giant Lab (JOGL) platform.

^cAPRICOT: Antigen Presentation Using Crispr for TB.

^dThese were projects that were not assessed by experts at the end of the program because they did not provide sufficient documentation.

^eThe Biloba project, which was not part of Co-Immune, was used as a base to create the team's own project, as the Biloba founder was a mentor during this event.

Co-Immune Project Assessment

Out of 22 projects, 7 (32%) provided sufficient documentation on JOGL to be assessed by the pool of independent experts. In total, 27 reviews were performed, yielding scores ranging from 18 to 32.8 out of a possible total of 45 across the different dimensions that were assessed (ie, approach, implementation strategy, and impact). The average score was 25.1 (SD 6.4).

HERA: A Health Platform for Refugees [53] was awarded with prizes, based on a total score of 15, for best approach (mean score 11.4, SD 2) and impact (mean score 14.6, SD 3.2). Commit to Get Vacc & to Promote Vaccination – HEROIC Santé [57] was awarded the best implementation strategy prize (mean score 10.33, SD 2.5).

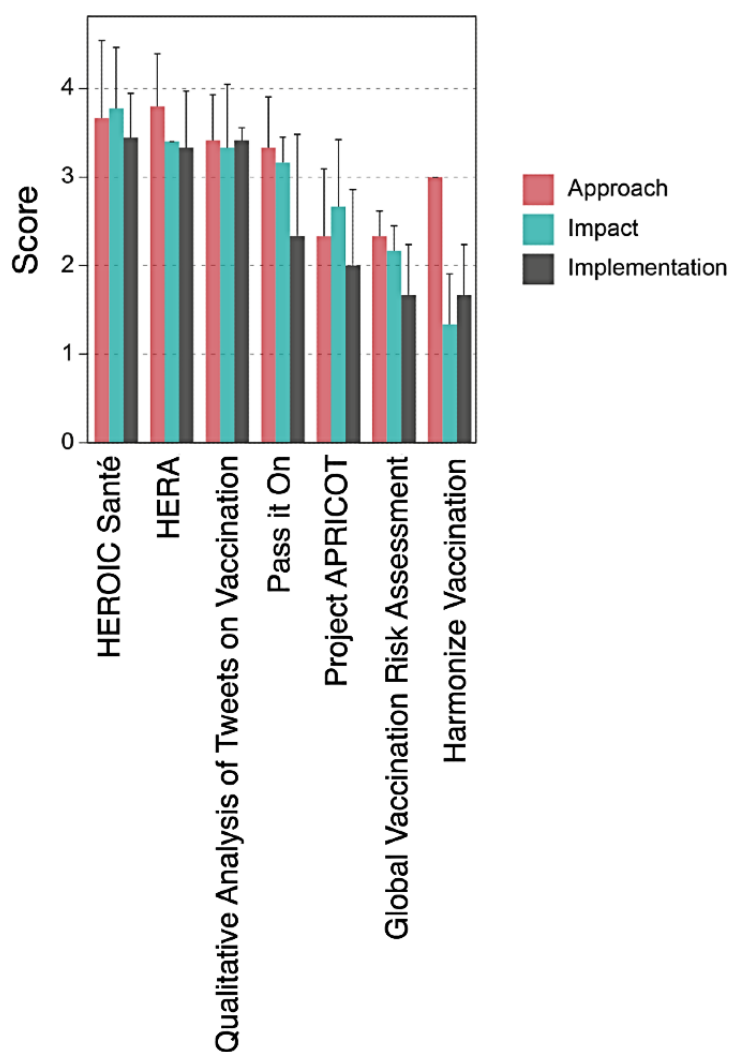
The projects were more successful, globally, in terms of approach, with a mean score of 9.37 (SD 1.79) out of 15 points. Out of 7 projects that were assessed, 4 (57%; Figure 8) had a score higher than 4 out of 5 for clarity, relevance, and alignment of their problem statement with the program objectives. For 6

projects (86%), the fit between the methods and the projects' objectives was scored highly by reviewers, with a score of at least 3 out of 5.

The implementation strategy score of projects was low, overall, given the early stage of the projects at the time of review. As such, only projects that existed prior to the program—HERA [52] and HEROIC Santé [57]—got a score of at least 3 out of 5.

For winners in each category, JOGL awarded them physical space for showcasing their project during the 2020 ChangeNOW forum at the Grand Palais in Paris as well as tickets for the Maddy Keynote, a major innovation event in Paris. Two partners—Excelya and the Wild Code School—also provided awards to the projects of their choice. Additionally, the Qualitative Analysis of Tweets on Vaccination [56] project was chosen to be the focus of a hackathon by the Wild Code School, and Project APRICOT [58] was offered technical support for data science and legal and regulatory affairs by Excelya.

Figure 8. Bar plot of review scores per category for all reviewed projects. Bars show average values for all questions related to each category, and error bars represent SDs. Projects are shown by decreasing global score. APRICOT: Antigen Presentation Using Crispr for TB; HERA: Health Recording App.



Discussion

Principal Findings

The Co-Immune program was designed to foster the creation and development of citizen science and open innovation projects addressing the contemporary challenges of vaccination in France and around the globe by reaching four specific objectives: (1) to foster collaborative, open, and transdisciplinary dynamics; (2) to promote the emergence of accessible knowledge and innovative solutions; (3) to support participants in the elaboration and development of their projects; and (4) to disseminate the outputs and results in an open science framework. Below, we discuss to what extent Co-Immune reached these objectives and highlight the challenges and facilitators in implementing such a program.

First, the program succeeded in creating a collaborative and transdisciplinary environment through its three core features: the JOGL platform, the organization of events, and the contest approach. This led to forming partnerships with 13 different organizations and recruiting over 230 participants, who displayed 492 unique skills and were engaged in creating 22 projects. The use of on-site hackathons was beneficial in

gathering nonacademic participants from various backgrounds. Our data show that in-person events and local outreach played a significant role in growing the community around Co-Immune. These offline events recruited 45% of the total community. Local enrollment was further strengthened by local partnerships, such as higher education organizations. However, the localization of our on-site events in Paris did not allow for the participation of people living in other parts of France or the rest of the world. Additionally, our online communication restricted the access of the online events to our realm of influence and to people with an internet connection. More inclusive participation geared toward people with diverse socioeconomic statuses and geographic situations is desirable in the future to give them agency over solving the problems that affect them. The development of new communities is usually a slow process in the absence of exogenous shocks, such as the surge in collaborative communities created by the COVID-19 pandemic [76]. Tapping into existing projects and networks for events has proven to be fruitful in our case, allowing for a steady growth of the Co-Immune community up until the end of the program. However, we did not observe further growth of the community after the end of the program. This highlights that in order to build a sustainable community using open innovation to tackle

global health challenges, one needs to facilitate the entry and exit of members, provide resources to support the current ones, focus on building on existing communities and projects, design inclusive environments for collaboration, and empower members to run their own activities.

Second, two design elements of the program converged to promote the emergence of knowledge and solutions to address aspects of access to vaccines and vaccination hesitancy: (1) the identification of challenges by experts in the field and (2) the alignment of the program strategy with national and international policies by frequent consultation with public health bodies and mobilization of members of public institutions in the CESI. Yet, greater representation of people affected by poor access to vaccines and people who are hesitant would be desirable to strengthen the alignment between the solutions developed and the most pressing needs at the local level.

Recently, online events have been used widely during the COVID-19 pandemic [76-78], supporting our initial assumption that forming and animating a distributed online community for public health programs is a relevant approach.

Third, the use of the JOGL platform, the mentorship during events, the assessment and feedback from experts, and the connection with a wide range of partners supported participants in the elaboration of their project in an efficient way. The use of the JOGL platform enabled projects to gain visibility, list their needs to create interfaces for collaboration, and share open data sets, code, and tools. Indeed, online platforms can offer projects that started at hackathons a pathway to pursue their development, potentially alleviating one of the main drawbacks of such short temporal interventions [43]. In this case, it also enabled the program coordinators to connect participants with project leaders based on a match between needs and skills. Yet, this approach was time-consuming, and scaling up our efforts proved to be challenging. The automation of such matchmaking tasks through a recommender system would help to minimize these efforts and increase the impact of projects through accelerated development [79]. In addition, mentoring is a known strategy that is used by open, online communities [80,81] and was leveraged by the Co-Immune program. Given the diversity of backgrounds and level of expertise across the participants, it was necessary to engage a similar diversity among the mentors. In our context, the highly rated projects that eventually received awards did not originate or participate in hackathons, but rather benefited from Co-Immune as a platform for further growth. Several of these projects already existed before the start of Co-Immune and had a higher maturity level than the projects created during the short span of the program. In addition, these projects were launched and run by people outside the larger Paris region. Thus, we stress the potential of online platforms and open innovation to build on existing projects and to replicate, adapt, and scale their activities in other contexts. Additional support consisted of promoting visibility on social

media by the organization team as well as opportunities for networking during events. Although no financial compensation was provided as part of this program, partners, through their own experts and co-organizing events, engaged in close relationships with JOGL and the individual projects. This was favorable for sustaining collaborations and projects after the end of the program. In the future, the sustainability of the newly created project efforts could potentially be improved by using incentives, such as microgrants or fellowship programs, for continuing projects in the postprogram period [79]. While the short time frame and limited resources allocated to the program did not allow us to implement a strong monitoring and evaluation strategy, future implementations should ensure that they conduct a minimum of pre- and postprogram data collection for assessing the full impact of the program.

Finally, the open science environment of this program was not only an asset for disseminating the outputs and results of the projects developed, but it also enabled them to replicate initiatives and, thereby, accelerate the resolution of the global health challenges they address. An example of this was given by the team from the project HERA: A Health Platform for Refugees [52], who opened its code, enabling any individual to replicate it. However, the lack of a thorough evaluation strategy prevents us from reaching a more definitive conclusion on the effective replication of projects carried out in Co-Immune.

Co-Immune showcases that short, focused programs can be efficient at mobilizing diverse communities in a rapid manner and harvesting ideas from various domains to address global health challenges. Yet, more case studies and evaluation work on similar programs are necessary to assess the full relevance of their design and the impact of the projects that are developed within them.

Conclusions

Co-Immune highlights how open innovation approaches and online platforms can help to gather and coordinate noninstitutional communities in a rapid, distributed, and global way toward solving SDG-related issues. The Co-Immune program gathered participants and partners from various backgrounds in a newly formed community to facilitate the creation of new projects as well as the continuation of existing projects to address the issues of vaccination hesitancy and access. In an open framework, the projects made their data, code, and solutions publicly available.

Through hackathons and other contest approaches, such initiatives can lead to the production and transfer of knowledge, creating novel solutions in the public health sector. The example of Co-Immune contributes to paving the way for organizations and individuals to collaboratively tackle future global challenges.

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Authors' Contributions

TL and IV co-designed the theme and the scope of the program at the early stage of the initiative. CM and TL conceived the program. CM led the coordination team of the program. CM, BGT, TL, CLBG, and MS participated in the program implementation. BGT, RJ, and MS analyzed data. CM, BGT, GF, CLBG, and MS wrote the paper.

Conflicts of Interest

GF was paid by Just One Giant Lab (JOGL) to support the elaboration of the manuscript; provided consulting services to Vaccines Europe, a trade association based in Belgium; and is a volunteer board member of the Coalition for Life-Course Immunisation, a UK-based charity. CM worked as the Co-Immune program coordinator for JOGL. BGT was an unpaid advisor to JOGL. JOGL received funding from Sanofi to develop and implement the program, which included publication fees. Sanofi respected the strict independence of JOGL, which administers its platform and the Co-Immune page in complete autonomy. Similarly, the Committee for Ethics, Science and Impact (CESI) was independent of Sanofi and decided alone on the strategic and scientific orientations of the program and the best projects to be rewarded.

Multimedia Appendix 1

Co-Immune partners and supplementary method for comparing Just One Giant Lab (JOGL) with other platforms.

[PDF File (Adobe PDF File), 196 KB - [jopm_v14i1e32125_app1.pdf](#)]

Multimedia Appendix 2

Co-Immune project assessment grid.

[PDF File (Adobe PDF File), 213 KB - [jopm_v14i1e32125_app2.pdf](#)]

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Abbreviations

APRICOT: Antigen Presentation Using Crispr for TB

CESI: Committee for Ethics, Science and Impact

CRI: Center for Research and Interdisciplinarity

HERA: Health Recording App

JOGL: Just One Giant Lab

SDG: Sustainable Development Goal

WHO: World Health Organization

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Original Paper

From Individuals to Systems and Contributions to Creations: Novel Framework for Mapping the Efforts of Individuals by Convening The Center of Health and Health Care

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Abstract

Background: People with lived health care experiences (often referred to as “patients”) are increasingly contributing to health care and are most effective when they are involved as partners who can contribute complementary knowledge alongside other stakeholders in health care.

Objective: Convening The Center aimed to bring together “people known as patients”—the center of health care—to address priorities as they defined them.

Methods: According to the original project design, an in-person gathering was to be conducted; however, as a result of the COVID-19 pandemic, the in-person gathering was transformed into a series of digital gatherings, including an in-depth interview phase, small-group gatherings, and a collective convening of 25 participants (22 women and 3 men from the United States, India, Costa Rica, Sweden, and Pakistan). Each participant was interviewed on Zoom (Zoom Video Communications Inc), and the interview data were thematically analyzed to design a subsequent small group and then full cohort Zoom sessions. Visual note-taking was used to reinforce a shared understanding of each individual- and group-level conversation.

Results: The interviews and gatherings for Convening The Center offered unique perspectives on patient activities in research, health innovation, and problem-solving. This project further developed a novel, two-spectrum framework for assessing different experiences that patients may have or seek to gain, based on what patients actually do, and different levels of patients’ involvement, ranging from individual to community to systemic involvement.

Conclusions: The descriptors of patients in academic literature typically focus on what health care providers think patients “are” rather than on what patients “do.” The primary result of this project is a framework for mapping what patients “do” and “where” they do their work along two spectra: from creating their own projects to contributing to work initiated by others and from working at levels ranging from individual to community to systems. A better understanding of these spectra may enable researchers to more effectively engage and leverage patient expertise in health care research and innovation.

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KEYWORDS

patient-centered care; patient role; patient involvement; access to care; patient-centered outcomes; co-design; participatory design; patient and public involvement

Introduction

People with lived health care experiences (often referred to as “patients”) are increasingly contributing to different areas of

health care, including research [1,2]. This strategy of involvement is most effective when individuals with lived experiences are involved as research partners [3] and contribute complementary knowledge alongside other types of stakeholders

[4], such as health care providers, purchasers, payers, policy makers, product makers, and principal investigators (PIs) [5]. However, these individuals are rarely presented with funded opportunities to directly connect with and learn from one another without the agenda-setting of sponsoring organizations [6,7]. To address this gap, the Convening The Center (CTC) project, funded by the Robert Wood Johnson Foundation, engaged 25 individuals representing diverse health conditions, geographies, and digital communities and provided resources to enable these individuals to gather and discuss the health care interests that mattered most to them—without influence from outside organizations. The needs of patient communities was the sole focus, and not an afterthought or engagement checkbox, of the CTC organizers (one of whom is a patient researcher herself and the other is a supportive collaborator). Embracing diversity improves patient outcomes [8]; however, diversity in patient participation in innovation remains a persistent challenge. Opportunities to participate in medical research and innovation mostly do not recognize conventional diversity criteria [9-12] or the talents and interests of patients; rather, patient participation in studies is often designed as a “check box” in funding proposals [13,14]. Archetypes of people with lived health care experiences have been defined in academic literature: the “difficult” patient [15], the “complex” patient [16], the “absent” patient [17], the “good” patient [18], or the “smart” patient [19,20]. These labels, not created by patients, fail to help researchers, health care practitioners, or other patients understand how to match the strengths, skills, and expertise of patients with participation opportunities.

This is the research gap that the CTC sought to better understand and explore. Many labels for types of patients exist; however, they are not used or adopted by the patients themselves, have not been collected into a cohesive map, and do not facilitate matching the skills and interests of patients with tangible opportunities for people with lived health care experiences to participate in improving the health care ecosystem at large.

The contributions of CTC are 2-fold: first, a patient-informed framework for mapping behaviors and activities for further study and exploration. The framework recognizes that the expertise patients bring ranges widely, including *creating* or initiating their own projects, communities, or solutions; *contributing* to other projects, research, or communities; articulating *individual lived experiences* (n=1) with particular health conditions; participating and engaging in *communities of patients* with differing lived experiences; and supplying expertise across multiple diagnoses, geographies, or digital communities at the *systems level*. Second, the methods used throughout CTC are novel and unique because they introduce participants to one another, establish trust, and facilitate conversations. This paper describes the methods of CTC, the range of topics that emerged from the cohort across the project’s discussions, the development of a novel framework for assessing patient experiences, and the potential applications of the framework for future use to improve the diversity of patient perspectives in research.

Methods

Overview

This formative, qualitative project used digital purposive sampling [21] of individuals with lived patient experiences. This involved a novel, three-phased approach: an initial phase to meet and develop relationships with individual participants, a second phase to engage small groups of participants to develop rapport within the cohort, and then a third and final phase to encourage deeper discussions.

Before phase 1, we sought to recruit a diverse selection of potential participants to CTC. The eligibility to participate in the project was broad; the project was open to anyone with new experience, a long history of working to improve health care through advocacy, innovation, design, research, or entrepreneurship, or other history of advocacy in a health-related domain. Initially, CTC was intended to be an in-person convening oriented to the priorities and interests of participants; travel costs and funding for participants’ time was outlined in the project budget, which was communicated to potential participants. This opportunity for the “periphery” of the health care space was intended to contrast with other health and health care events sponsored by the “center” of the health care space [22]—companies, insurers, provider networks, research funders, or academic societies—which naturally focus on issues of concern to those stakeholders rather than on the needs and quality of life of people with lived health care experiences. After the COVID-19 pandemic disrupted the plan for individuals to meet in person, CTC was redesigned as a digital activity. The reduction of travel and venue costs enabled an increase in participant honoraria. This change was announced to the initially nominated participants, and additional time was added to this stage of recruitment.

Participant recruitment was conducted in multiple stages to enable both self-nominations and nominations of others who may not have been aware of the potential opportunity, being outside of the one to two degree-connections of the research team.

For initial nominations and recruitment, a Google Form was created ([Multimedia Appendix 1](#)) and shared through a blog post on the PI’s blog (primarily about lived experiences with multiple chronic conditions) [23] and the investigators’ Twitter accounts [24], with requests for interested participants to further share the form with additional patient communities. To increase the research team’s ability to reach different communities, the form asked for additional communities and organizations that should be notified of the CTC program. After the first Google Form was closed, 90 nominees were contacted via email with invitations to complete a second Google Form ([Multimedia Appendix 2](#)) with additional demographics and a response to the question “What inspired you to want to make a difference in health care?”

The CTC PI (DL) and co-PI (JH) thoroughly reviewed all applications on an ongoing basis to ensure that a mix of individuals had been nominated to represent rural and urban settings; a diversity of ages, geographies, races, and ethnicities;

and various gender orientations. After the application period closed, the investigators (DL and JH) applied diversity criteria to select a cohort that prioritized the participation of individuals who were: Black, Indigenous, or People of Color; women; and residents of rural areas. Diversity in the participants' experience in research was also considered. A total of 41 individuals completed the second stage of nomination, and ultimately, 25 individuals were selected to participate. This cohort size was partly driven by the original budget and plans for an in-person gathering, where up to 30 US-based individuals would have been selected to participate in person. Cohort size was shaped by the research team's availability for facilitation, the dynamics of group sizes in digital meetings [25], the time and resources necessary to support visual note-taking, and investigator assessment of how individuals were positioned to uniquely contribute to and benefit from participation in the cohort.

The 25 individuals were chosen for the cohort primarily based on their responses to open-ended questions about their work or interests in the health spaces that led them to apply for CTC. However, after the initial selection, based on the open-ended answer content, the research team reviewed additional metrics to ensure that they did not repeat the structural biases that may have influenced how individuals responded to the questions in the application for nomination. The final cohort was consequently diverse across several metrics. We asked nominees how long they had been advocating in the space they had described in their application: 8% (2/25) reported 1 to 2 years, 20% (5/25) reported 3 to 5 years, 40% (10/25) reported >5 years, and 32% (8/25) chose the option of "It's complicated to answer - I've been working on multiple problems over time." When asked for information on race, 64% (16/25) reported White, 20% (5/25) reported Asian, 8% (2/25) reported Black or African American, 4% (1/25) reported Hispanic or Latino, and 4% (1/25) reported American Indian or Latina. Age was the most balanced metric out of those that we evaluated: 20% (5/25) were aged 25 to 34 years, 28% (7/25) were aged 35 to 44 years, 24% (6/25) were aged 45 to 54 years, 20% (5/25) were aged 55 to 64 years, and 8% (2/25) were aged >65 years. Gender was the most imbalanced metric in the final cohort: 88% (22/25) of participants were women. After a deep discussion evaluating additional individuals who had identified as men based on the fit with the rest of the cohort, we ultimately did not expand the cohort to additional men participants based on gender imbalance, as we weighted lived experience higher as a criterion than attempting to increase men in the cohort. The cohort represented

lived experiences across numerous areas, such as disease-specific communities (eg, lung cancer, breast cancer, diabetes, and various rare diseases), as well as cross-community topics (eg, trauma resulting from or related to health care and a focus on diversity, equity, and inclusion in advocacy spaces). The cohort was primarily based in the United States (19/25, 76%) but also included participants from Costa Rica (1/25, 4%), Sweden (1/25, 4%), India (3/25, 12%), and Pakistan (1/25, 4%).

To begin building rapport with the selected cohort, the investigators designed an informal, semistructured [26] interview protocol (Textbox 1). The goal of these conversations was to listen deeply and understand the perspectives of each participant, including changes in their efforts during the COVID-19 pandemic. The investigators interviewed each of the 25 CTC participants one-on-one through Zoom, which constitutes phase 1.

Two notetakers were present during the phase 1 Zoom calls. The first was the PI (DL). The second was RR, who kept their camera and audio off throughout phase 1 and was introduced as an additional notetaker. The lead interviewer during phase 1 was co-PI JH. This was intentional, as PI DL had existing relationships with a few participants; others may have had name recognition or awareness of PI DL's own work in this space, and it was possible that these factors may influence discussions. PI DL self-assigned herself to a note-taking role to minimize her influence on the direction of these initial conversations. At the end of the call, PI DL was invited by co-PI JH to re-enter the conversation to help answer questions from the participants about the next steps for the project and what to expect.

Unbeknownst to each interviewee, the second notetaker during phase 1 (RR) was an artist assigned to develop a "visual note"—an illustration—of each participant's conversation. The research team used follow-up calls to present each participant their visual illustration, which was intended to be a gift to the participants that they could use in the future. During this second Zoom call, the research team presented the artwork, sought initial reactions to it, and asked the participants to request changes or edits to the visual notes to ensure that their gift accurately portrayed their involvement and experience in addition to how they preferred the art piece to appear. This helped the research team represent participant experiences as intended and aligned the artwork with the purposes for which the participants might use it.

Textbox 1. Semistructured interview questions used in Convening The Center.

Intent and the corresponding interview questions

- General introduction questions and seeking to understand their efforts and problem spaces
 - How are you doing?
 - In general, how comfortable are you talking with others about your personal health or your personal story?
 - What did you have to do to prep for this call?
 - Tell me about yourself and how you found yourself working to fix something in health care.
 - How long have you been in the health care–fixing space?
- Addressing the elephant in the room: the pandemic
 - Tell me about your biggest consistent challenges during the pandemic.
 - If you transport yourself back to 2019, in the fall, before the pandemic, can you remember what your biggest challenges were then?
 - Are there any pandemic-driven changes that you appreciate? What pandemic-driven change has been the best for you?
 - What do you predict the biggest challenges will be in your space after the pandemic?
- Discussing any differences in how participants and their works are seen from different perspectives
 - How do you think that others (physicians, friends, family, and public) see you?
 - How would they describe you or your work?
 - How would you like others to see you?
 - How do you see yourself?
 - Are you familiar with the term “imposter syndrome”? Have you ever experienced imposter syndrome? Tell me about how you felt and why you felt so.
- Learning about the types of activities
 - Where do you feel like most of your patient-or caregiver-or advocacy-related time is spent?
 - What type of activities do you find yourself doing most?
 - Where would you like to see most of your time spent in the future?
- Learning about the skills they use and would recommend to others
 - What is the most important skill set for a new patient advocate to have? Why that one?
 - What one skill set do you have that you would give to a new fellow advocate or doer?
- Systems-level questions about skills and efforts that might translate to different communities
 - Of your work in this space, what do you think might translate to other patient communities or other health care spaces? Why?
 - Which work would not translate? Why?
 - What have you absorbed or translated from another patient community or space that you have found useful?
- Learning about the ideal design of digital gathering and to inform the design of phases 2 and 3 within the project
 - Tell me about the worst digital event or experience you have had during the pandemic.
 - Tell me about the best digital event you have attended during the pandemic.
 - Tell me about the best digital community you have been part of.

The goals of providing visual notes to the CTC participants as part of phase 1 were to achieve the following:

1. “Surprise and delight” [27] participants and signal the intent that CTC was an experience beyond what patient advocates may have come to expect from the research participation process
2. Demonstrate the commitment of material resources made to the cohort, who are often not resourced for their work
3. Show that the research team listened to and heard each participant’s individual perspectives
4. Visualize each participant’s story as an artifact under their control and with probable personal and professional

applications (eg, conference talk introductions or sharing the CTC experience with family members)

After phase 1, CTC participants were invited through email to join a workspace on Slack (Slack Technologies), a digital collaboration and chat platform. Although participation on Slack was not mandatory, many joined the platform to meet one another, and many participants shared their visual notes to introduce themselves.

To prepare for phase 2, the investigators thematically analyzed the collected interview data [28]. Phase 2 consisted of 4 small-group Zoom meetings, with up to 8 participants. Groups were selected based on a mix of availability and personality to ensure to the best of the research team's understanding of personalities that each phase 2 conversation would allow space for all voices to contribute.

Unlike phase 1—for which there was an interview protocol—no formal agenda was set for the phase 2 calls, which was core to the project's purpose of bringing individual patients together without an agenda. To help put each group at ease, a Google Slides deck was created to visually anchor conversations, and a link to editable slides was shared with the group during Zoom calls (Multimedia Appendix 3). The slide deck included introductory (icebreaker) activities [29] to help participants introduce themselves and their work and highlighted shared interests and themes that participants might discuss, as well as introduced them to the tool used for group note-taking. Visual note-taking took place for all 4 groups during phase 2, and participants were informed that it would help summarize the group discussions. Similar to phase 1, after each session, the visual note was presented back to the group, and participants were invited to suggest edits or changes to the visual note reflecting the conversation in an attempt to continue to promote a shared understanding of the experience.

After phase 2, the research team began mapping the participants to better understand the emerging perspectives among the cohort.

The perspective maps were plotted on a 2D scale, which the team called the Two-Spectrum Assessment of Patient Experience (further discussed in the Results section). From this visual plot of participants, the research team identified groupings of participants with similar experiences within the cohort, and these groupings were later used to determine the makeup of smaller groups during breakout discussions in phase 3. Using Slack, participants were surveyed about phase 3 discussion topics emerging from phases 1 and 2.

In phase 3, all 25 CTC participants gathered for a 2-hour Zoom call. Similar to phase 2, Google Slides was used to facilitate icebreaker activities as participants joined the call (Multimedia Appendix 4). After the icebreaker activities, 3 rounds of breakout discussions took place: (1) introductions, (2) affinity groups, and (3) topic-based groups. These groups have been described in detail in Textbox 2.

After the third breakout session, there was a short break, and then all the participants returned to the main Zoom room where an agenda-less discussion took place among all 25 participants. Following phase 3, the final visual note reflecting this penultimate cohort conversation was shared, and the participants were again encouraged to provide feedback, including suggested edits or changes to further adapt the visual note.

To summarize these methods, phase 1 participants were individually engaged to get acquainted with them at an individual level and develop rapport with the research team, with the goal to surprise and delight participants through their participation in this project. As for phase 2 participants, we sought to develop rapport within small groups in the overall cohort and begin to foster ideas for the subsequent agenda-less conversation. Finally, in phase 3, we intended to design a meaningful agenda-less gathering in the spirit of the original project design while providing enough structure and support to catalyze the conversation among participants. Visual note-taking was used to advance a shared understanding of the discussions at the individual and group levels for each set of conversations.

Textbox 2. Groups of breakout discussions in phase 3 of Convening The Center.

Breakout groups

1. Introductions

- The goal of the first breakout group was to introduce the participants to fellow participants whom they had not met in the previous phase 2 small-group interactions or had not previously known outside of Convening The Center. This was an opportunity for the participants to get to know the other members of the cohort better, and there was no formal agenda.

2. Affinity groups

- The second breakout was designed around affinity groups of participants who were working at similar levels (eg, individual solutions vs community), had similar experiences (eg, working in breast cancer communities), or had expressed interest in similar future directions (eg, patient-led research). These small groupings were determined by the research team when reviewing the visual plotting of participants on the Two-Spectrum Assessment of Patient Experience and chosen following phase 2 discussions. The small groupings included participants who were newer or getting restarted, those experienced at the community level regardless of topic or space, those who were creators or initiators of projects and communities, and those with experience at the systems level across multiple communities.

3. Topic-based groups

- The third round of breakouts was based on topics that participants had identified and voted on in Slack or by email after phase 2. In this grouping, individuals joined breakout rooms based on the topics that were most interesting to them.

Patient and Public Involvement

Patients and the public were involved with CTC at every stage, starting from its inception. This was a patient-led (DL) project, and a compensated advisory group of 2 experienced patient advocates (LS and AS) also contributed to the overall project design and recruitment strategy and helped guide the project's development.

All participants were paid US \$1000 for their time spent on participating in all 3 phases of the project: 6 hours of synchronous gatherings between phase 1 (90-minute initial conversation and 30-minute follow-up conversations), phase 2 (2 h), and phase 3 (2 h). Participants were offered the opportunity to donate their compensation to a nonprofit organization of their choice or be paid directly for their time. For some of the cohort, this was the first time that they were offered any form of compensation for their contributions as patient advocates.

Participants were given opportunities to provide feedback on the project throughout the 3 phases, including feedback on their individual artwork, preferences for the design of digital gatherings (phase 1), group visual note feedback (phase 2), and topics for breakout groups (phase 3). Investigators asked for and received feedback from the participants immediately following the project, as well as on the draft report to the funder Robert Wood Johnson Foundation, resulting in additional content for the report [30]. Similarly, this manuscript was presented in a draft form to the cohort, seeking voluntary feedback and input from the participants who chose to review it, and their input was included in this manuscript. CTC participants are recognized in the Acknowledgments section of this paper.

Ethical Considerations

Because this project was designed to be a participatory project and not "research," human participant approval or ethics approval was not sought in accordance with the 2018 Common Rule 45 CFR §46.102 (1)(1) of the US Department of Health and Human Services, Office for Human Research Protections [31]. The process of consent was opt-in to participate in the project at each phase, which included disabled camera and audio until a participant chose to enable (turn on) audio or video based on their individual choice. Participants were encouraged and reminded that they could turn off their video after they turned it on (if they had turned it on previously) for any reason. Once the investigator team realized that there was content worth sharing outside the project about the methods of the project and the high-level discussion themes raised by the participants, the idea of generating a paper was brought to notice of the CTC participants. As described above, the participants were invited

to participate, and many provided direct input to this article. The original intent of the paper was to present the artifact of the Two-Spectrum Assessment of Patient Experiences, which has been described in further detail below and expanded upon reviewer feedback to also include elements of the novel design methods used to design the project itself.

Results

Overview

CTC was not conceptualized, developed, or implemented with the intention to make it an academic research project; rather, it was designed as an opportunity for people with lived health experiences (patients) to produce relevant knowledge about how patients participate in health care research and innovation. This paper reports on "patient knowledge," which has the potential to frame research activities to better match opportunities with the individuals who would excel at them, and offers a summary of the thematic discussion topics that arose when CTC participants were encouraged to gather and set their own agenda.

Two-Spectrum Assessment of Patient Experience

The primary research result from CTC was the development of a *Two-Spectrum Assessment of Patient Experience* (Figure 1).

The horizontal spectrum represents the *types* of involvement: whether someone is typically participating as a *contributor* to another project or effort, led by someone else, or are serving as a *creator* of the project or community themselves. The horizontal spectrum is "contributor" to "creator." The vertical spectrum represents the *scale* of involvement. Level 1 indicates an *individual* level of involvement. Individuals typically start here as patients, where they identify problems or opportunities to improve things for their personal journey as a patient. Level 2 indicates a community level of involvement, that is, involvement in a community of any size, such as a disease-based community (eg, diabetes community), a specific geographic community (eg, rural Appalachia patients), or a digital community (eg, patient-run support group on Facebook). Level 3 indicates a systems level of involvement, meaning that the work transcends multiple specific communities and likely impacts multiple communities, disease spaces, or areas of health care (eg, working to improve access to electronic health records).

Figure 1 illustrates this grid as used within the project, with boxes representing individual participants based on researcher assessments of the participants' articulation of their work during phase 1 interviews. (Figure 2 contains a blank grid for visualizing how this might be used by others in the future).

Figure 1. An example of the two-spectrum patient experience grid, with 25 individuals from Convening The Center mapped on the grid as assessed by the project investigators.

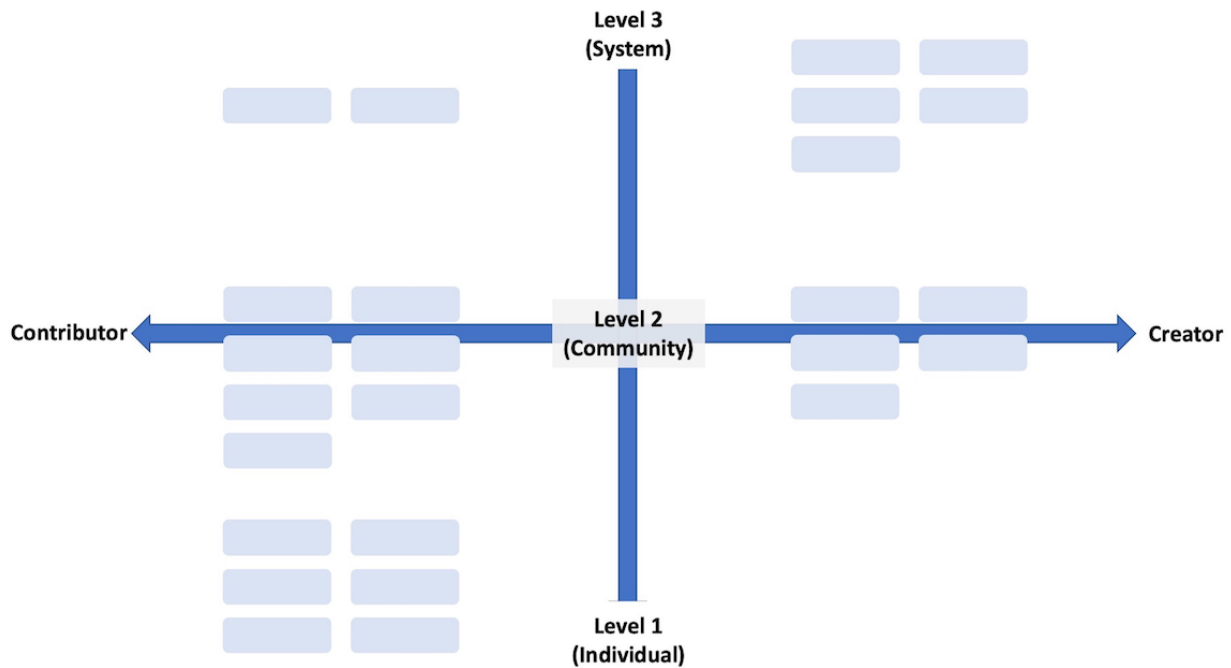
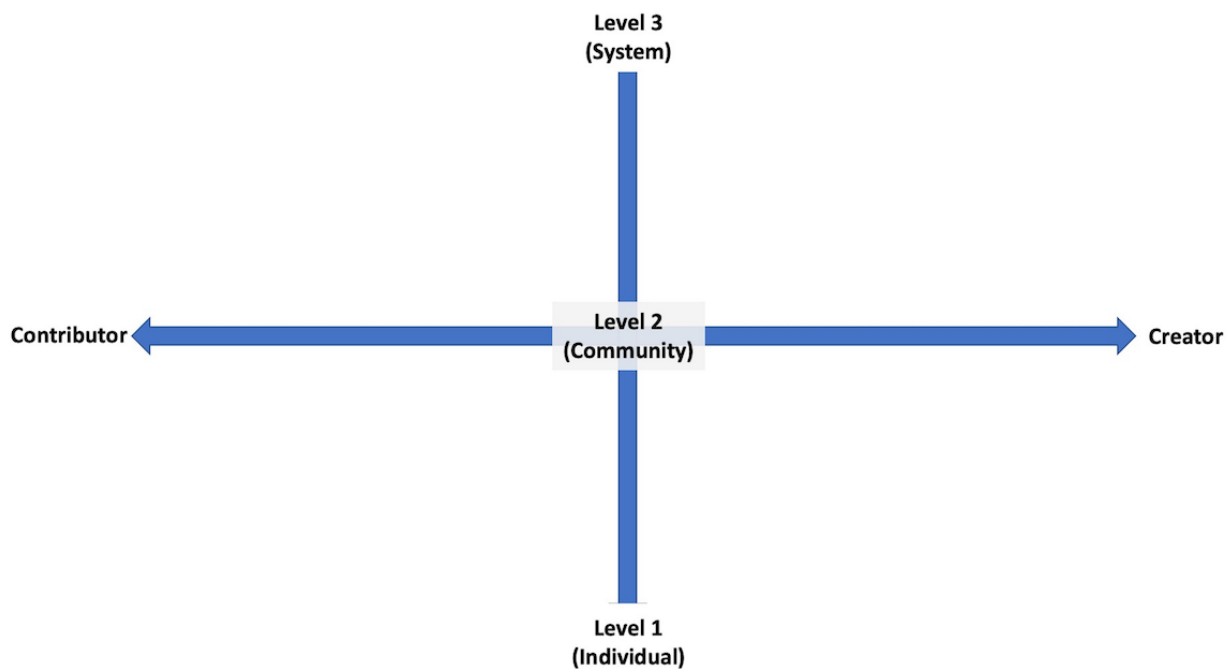


Figure 2. Illustrates a blank version of the two-spectrum assessment of patient experience grid as used within the project, with boxes representing individual participants based on researcher assessments of the participants’ articulation of their work during phase 1 interviews.



Recurring Themes of Discussion by Participants

During phase 2 small-group discussions, some themes reoccurred independently across the groups and were voted on to be included in phase 3: research, identity, medical education and working with health care providers, and mental health.

In the resulting phase 3 discussions, the topic of *research* focused on the opportunities and experiences of patient

researchers themselves (eg, to become a working member of a research team or taking on a leadership role such as a coinvestigator). These discussions covered self-directed research, and the research participants contributed to and highlighted research dissemination strategies, such as through traditional medical journals and social media.

The discussion on *identity* centered on the perspectives of being a “patient” or “carer” and identity labels (eg, diagnosis, gender,

Discussion

Principal Findings

This novel, patient-led project sought to fund a gathering of diverse individuals seeking to improve health care outside of the traditional avenues of working professionally in legacy health care institutions. As a result of the COVID-19 pandemic, CTC pivoted focus to a digital series of synchronous and asynchronous gatherings to achieve the goal of bringing together a diverse cohort of patient advocates and fostering a sense of trust, openness, and community to facilitate discussions based on the interests of the cohort. As a mechanism for grouping smaller conversations within the cohort, a two-spectrum framework of patient experience was developed to articulate differences and similarities outside of typical criteria such as gender, race, geography, or disease and provide opportunities to reflect on the contributions to or creations of existing or novel efforts as well as individual-, community-, and systems-level experiences. This two-spectrum framework does not define patients as “good” or “difficult”—as seen in the academic literature sometimes—nor does it categorize patients along demographic criteria such as race, gender, geography, or disease. *Rather than making claims about what patients “are,” this framework describes what patients “do,” the often-unseen work of patients, and, importantly, how they do this work.* By better understanding and mapping what patients “do,” there is a great potential to ripple outward from the center of health care (patients) and influence future innovation throughout health care spaces.

This framework was developed to aid the research team in understanding the clusters of alignment and differences within the CTC’s 25-member cohort; however, we realized that this framework could also be useful outside of this project. First, patients may be able to use the *Two-Spectrum Assessment of The Patient Experience* grid to self-identify *where they are* currently in their experiences and interests. Second, they may be able to identify a *direction* in which they would like to head, “see” other patients in those areas, and seek mentorship, partnership, or support from those individuals. They may also use this framework to assess invitations and opportunities to contribute to traditional research projects or health care improvement initiatives.

It is important to note that not all patients intend to be involved in any particular place in this matrix. There is no right or wrong place to be or strive to be. However, the matrix can be used by individuals with lived health care experiences to self-assess their current and prospective work. It can also aid patients in assessing opportunities to determine whether they match the type of work that they want to do. Similarly, researchers, organizations, companies, and others can use this framework to better articulate and define what level of involvement and participation they are striving for when seeking patient involvement in a project or research initiative.

This framework may help all sides better articulate the expectations of what a project entails and determine whether an individual is an ideal fit for a project and importantly,

whether the patient advocate is even interested in the project being presented by a funder or other entity.

For researchers, organizations, and others seeking to involve patients and members of the public in their work (eg, patient and public involvement programs), the *Two-Spectrum Assessment of Patient Experience* grid could aid in recognizing that patients are not “one size fits all.” Different patient advocates have a variety of life experiences, interests, and backgrounds from previous work, and projects may need varying levels of participation and expertise. As such, the Two-Spectrum Assessment of Patient Experience may be used to matching opportunities with the right person rather than assuming that “any patient will do” to check a box of patient involvement.

Moving forward, this framework can be used as a tool to support the increasing involvement and resourcing of patients who are seeking or find themselves facing opportunities to help fix the parts of health care that are not working for them or their communities. It can also be used to help researchers assess blind spots to identify where they may be missing additional patient expertise and potentially creating inadvertent sources of bias. It is a potential tool to help improve the relationship between traditional researchers and the invited patient contributors to research or other health care improvement initiatives (eg, the authors also built the “Opening Pathways Readiness Quiz,” intended to assess researcher readiness to collaborate thoughtfully with patients [32]). Further tools should be developed to connect patients with varied experiences and interests to research, as well as to help patients assess their fit and interest in individual opportunities.

Beyond the framework, the key themes identified in CTC that recurred across discussions of all sizes (individual, small group, and larger cohort) are worth noting for those interested in encouraging and expanding further patient involvement and engagement in research, advocacy, and other areas of health care improvement. The themes collectively reflect both avenues of opportunity—such as self-research or contributing or creating new research or contributing to medical education—as well as challenges that participants (“patients”) experience, such as grappling with their own identity or the identity of the communities in which they participate, and the mental health aspects of being involved above and beyond one’s own health care.

In addition to the recognition of the framework as a tool to assess people as multidimensional, traditional researchers should be aware that patients are unique individuals while still sometimes sharing similar challenges across different types of communities (eg, geographic or disease). Participants in CTC, especially those with a longer duration of experience, frequently remarked on observing efforts “recreating the wheel” in those newly participating. This applies to both those with lived experience as well as traditional researchers: both groups should look for examples or inspiration not only within the disease or health area on which they are focusing but also within other groups or disease spaces [33]. For example, a rare disease community may benefit from another disease community’s efforts. Traditional researchers may benefit from sharing expertise and experience for building partnerships with

individuals with lived experiences. Just as cross-disciplinary collaboration in traditional research can find applications of existing solutions in new areas, cross-disease and cross-community collaborations may also inspire reuse of solutions or further innovations to solve some of the unsolved challenges that may exist in another area.

Participants in CTC expressed the desire to continue to expand their reach and impact, perhaps with new collaborations or partnerships. One idea worthy of further exploration is platforms or opportunities to “benefit all stakeholders,” which would better connect funders, traditional researchers, and organizations or institutions with patients and existing community networks. There is an awareness within this participant group of survivor bias [34] and the privileges that some participants experience or have experienced that have led to the opportunities for their contributions in these spaces. Awareness of this should also be raised and brought to the attention of traditional researchers when assessing potential partnerships with those with lived experiences.

It is worth highlighting the conscious design efforts that went into the CTC gatherings to facilitate discussions. It was not simply another digital meeting. The 3-phase design was orchestrated to bring together a group of total strangers without an agenda, which provided challenges for an in-person or digital gathering. We chose to design sessions for individuals to small groups to an entire group discussion but further broke up the all-cohort gathering into several parts to continue to grow relationships, build comfort and trust, and provide space and methods for people to communicate their ideas and interests. We invited contributions through speaking, writing through the Zoom chat functionality, and writing notes shared on Google Slides throughout the gatherings ([Multimedia Appendix 4](#)). We specifically invited contributions and edits to the visual note prepared in all three phases. This contributed to a shared understanding of the content discussed, ensured topics or moments that resonated with participants were reflected in the notes or output of the discussions, and attempted to extend the partnership of the participants in the project.

For future meetings that bring together participants with lived experiences as advocates or research partners, or in combination with traditional researchers as partners, we encourage further thought and consideration of the inclusive design of digital gatherings. Although we by no means are experts, our consideration list for this meeting and future meetings involves:

conscious choice of meeting platforms that work for all participants; flexibility in whether video is “required,” for both internet bandwidth and participant energy bandwidth purposes; including breaks and being cognizant of the length of gatherings; and designing input methods to consider personality types (eg, introverts vs extroverts or different communication styles) or those who prefer written over verbal communication.

Visual note-taking played a more impactful role than expected for the research team, in addition to meeting our intended goal of “surprising and delighting” participants by gifting them with the work of art to reflect their stories and experiences as shared with us in phase 1. Visual note-taking and our methods of asking participants for edits or changes at each phase advanced shared understanding not only for the participants but also for us as organizers and researchers. The visual notetaker did not have any biases or experiences that the investigators (DL and JH) had in health care and lived experience spaces, providing a fresh perspective and neutral “ears” to each conversation, which contributed additional findings to the research team after reflecting on each discussion and each phase of the project. Using visual note-taking to support written or transcription notes for meetings or significant gatherings is a method we would recommend others use, preferably in the digital format or otherwise in a way that invites two-way contributions to highlight any missed content or opportunities to edit to ensure accuracy.

Conclusions

CTC was a patient-led initiative funded and organized as a gathering of diverse individuals working to improve health care outside of more traditional frameworks (eg, working professionally in legacy health care institutions). Gathering digitally, the participants highlighted some of the complexities and challenges of working to change health and health care from the outside, while also highlighting similarities in efforts across different communities. In contrast to the academic literature labeling what patients “are,” the CTC *Two-Spectrum Assessment of Patient Experience* framework describes what patients “do” when they go beyond navigating their individual lived health care experiences and transition toward community- or systemic-level involvement. Better understanding and mapping what patients “do” has the potential to ripple outward from the center of health care (patients) and influence future innovation throughout health and health care spaces.

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Conflicts of Interest

All authors have completed the International Committee of Medical Journal Editors uniform disclosure [35] and declare the following: all authors received financial support from the Robert Wood Johnson Foundation for the submitted work; LS reports grant funding from the Gordon and Betty Moore Foundation, Cambia Health Foundation, The John A Hartford Foundation, Agency for Healthcare Quality and Research, and the National Cancer Institute within the previous 3 years; and no other relationships or activities that could appear to have influenced the submitted work.

Multimedia Appendix 1

Initial nomination form for Convening The Center.

[PDF File (Adobe PDF File), 78 KB - [jopm_v14i1e39339_app1.pdf](#)]

Multimedia Appendix 2

Second nomination form for Convening The Center.

[PDF File (Adobe PDF File), 232 KB - [jopm_v14i1e39339_app2.pdf](#)]

Multimedia Appendix 3

Slides used in phase 2 small-group conversations for Convening The Center, including examples of interactive components for introducing the participants to editing the collaborative slide deck.

[PDF File (Adobe PDF File), 767 KB - [jopm_v14i1e39339_app3.pdf](#)]

Multimedia Appendix 4

Slides used in phase 3 of Convening The Center, with examples of interactive components for introducing the participants to editing the collaborative slide deck and displaying the overall agenda and flow for the all-cohort conversation.

[PDF File (Adobe PDF File), 91 KB - [jopm_v14i1e39339_app4.pdf](#)]

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Abbreviations

CTC: Convening The Center

PI: principal investigator

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Original Paper

A Norm-Creative Method for Co-constructing Personas With Children With Disabilities: Multiphase Design Study

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Abstract

Background: An increase in the demand for child participation in health care requires tools that enable and empower children to be involved in the co-production of their own care. The development of such tools should involve children, but participatory design and research with children have challenges, in particular, when involving children with disabilities where a low level of participation is the norm. Norm-creative and participatory approaches may bring more effective design solutions for this group. “Personas” is a methodology for increasing user perspectives in design and offers representation when users are absent. However, research on participatory persona generation in this context is limited.

Objective: The objective of this study was to investigate how norm-creative and participatory design approaches can be integrated in a persona generation method to suit children with disabilities in the design of games for health that target this group.

Methods: The method development involved interview transcripts and image-based workshops. Sixteen children with various disabilities participated in persona generation through co-creation of characters and scenarios. The results from the workshops were validated together with 8 children without disabilities, 1 young adult with a disability, and 1 rehabilitation professional. A qualitative thematic design analysis was iterated throughout the process.

Results: The results consisted of an image-based and iterative co-construction method. It was accompanied by examples of personas that were generated and validated within a games for health case. The method showed effectiveness in enabling flexible co-construction and communication. The data resonated with social model perspectives, and the development is discussed in terms of participation levels, salutogenic descriptions of barriers, and norm-creative tradeoffs.

Conclusions: The resulting method may influence future design projects toward more inclusiveness and enable increased representation for children with disabilities in research and design. Using this method to its full potential requires a norm-critical awareness as well as extensive facilitation. Suggestions for further research include the application of the method to design processes in similar contexts or user groups.

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KEYWORDS

disability; children; norm-critical; participatory design; personas; co-produced care; health care

Introduction

Background

Recent research has shown that while participation is on the agenda of health care professionals, practical guidelines and tools for child participation are lacking [1-5]. This affects

children whose lifestyles involve a close relationship with health care services. The pursuit of increased participation for children should also cover research and design processes where children, and particularly children with disabilities, are often left out [6-9]. Child participation is fundamental to ensure a user perspective and to increase the chances for the successful design

and implementation of new solutions for children. However, participatory research with children, and especially children with disabilities, has challenges. Since this group has a broad range of special needs, participatory processes depend on customized and user-centered methods [8,10-12]. Overcoming barriers for involvement related to age and disability requires a norm-critical mindset. This allows for a rethinking of power distributions and conceptions of disabilities as barriers. This study thus uses a norm-critical approach to investigate how participatory design methods can enable and capture children's perspectives to inform design processes that target children with disabilities.

Co-produced Care: Benefits, Barriers, and Risks

The concept of health care as *co-produced*, as opposed to simply delivered, aims to increase both the quality and efficiency of care [13,14]. Participation is a prerequisite for co-production and has the potential to increase patient empowerment [15,16], motivation, and the effectiveness of interventions [13,17,18]. The benefits of participation for the quality of care have also been found in pediatric contexts. These include better preparation, greater control, feelings of self-esteem, less anxiety, and fewer risks [4,15,19-23]. Research has also shown that children wish to be more involved than they are [24]. Participation is therefore generally regarded as a priority and a prerequisite for good care by health care professionals [4,25]. The *human right* to participate as advocated by United Nations' Convention on the Rights of the Child [26] thus aligns with this ongoing paradigm shift in health care. Co-produced care channels more control and responsibility over the care processes toward the patient [13,14]. Following this logic, the lesser the patient participates, the greater the risk of lower quality of care. Patients facing barriers for participation thereby risk becoming even more marginalized.

Children with disabilities represent a group that might not manage to fulfil the requirements or norms of co-production unless health care services are designed appropriately. In addition to the patient-professional hierarchy, barriers to co-production are inherent in being a child in an adult domain, such as through parental gatekeeping, communicative inequalities, professionals' resistance toward power sharing, and a disbelief in children's capabilities [23]. Despite an awareness of the benefits of child participation, professionals largely fail to achieve this in practice, commonly manifested in failing to address the child [4,23,27]. Moreover, pediatric health care uses a *family-centered approach* in many cases, that has been criticized for blurring the boundaries of who is the real client and risking that the child's perspective is not prioritized [7,28]. Recent research has thus highlighted the importance of child-centeredness in pediatrics to increase the safety, quality, and perceived value of care [3,5,27-30]. Increasing child-centeredness while at the same time reducing an established family focus must involve additional support for children, since participation requires both involvement and responsibilities. Research-based design methods and tools that empower child patients to independently advocate their own needs and preferences could create such support. One digital category of tools is *games for health* (also called serious games for health [31]). Games for health motivate patients through

characteristics borrowed from entertainment games but with health objectives such as maintaining, restoring, and personalizing health [32].

Involving Children in Design: Participatory Approaches and Norm-Critical Perspectives

The design of tools that aim to increase the participation of children in their own health care needs to be based on the involvement of children from the specific target group. *Participatory design* is increasingly being used to deal with social and health-related issues as it offers a range of creative methods to promote user perspectives [16,33,34]. Participatory approaches might, however, need to be adapted to allow for the inclusion of groups that are too marginalized to get involved through conventional health care fora [10,12,35].

Critical perspectives have come to be influential within this field. Norm-critical and social models of disability are both rooted in critical theory and treat normality, functionality, and disability as malleable and context-bound [36,37]. Norm-criticism aims to identify and question excluding norms within a given context. This involves shifting the focus from a disability-oriented *pathogenic* focus toward a resource-oriented *salutogenic* focus within health care. Salutogenesis acknowledges an individual's goals, preferences, and resources as keys when working toward better health [38]. Norm-criticism can expose norms and their mechanisms, which in turn can be used as a springboard for *norm-creative* solutions that serve to remodel norms in a direction of empowering practices [37,39,40]. A combined approach based on norm-critical and salutogenic perspectives is thus favorable when seeking to create solutions beyond excluding norms (ie, norm-creative solutions). Unlike explicit norm-critical design, this study thus uses norm criticism as a means and not an end to reach usability and impact. A critical approach is also useful when involving marginalized groups in research and design contexts, in order to challenge power hierarchies and perceptions of barriers [37]. Given that misassumptions about child users are more common than in designs for adults [41,42], this approach relates to the process of identifying and bringing forward children's preferences and interests in the design process. User participation is necessary to understand user needs [42,43], and a corner-stone for participatory design is to realize that most people can contribute to creative processes when given the right support [11,16,44,45]. Although vulnerable user groups, such as child patients, are becoming increasingly involved in participatory design within games for health [43], such involvement is far from easy.

Challenges in participatory design with children in health care contexts include recruitment (since child patients often have limited energy and spare time) [46], extensive preparations [47,48], and time-consuming data collection (partly since children have more difficulties verbalizing abstract concepts and actions) [27,41,47]. All the above are particularly true for children with disabilities. In addition, when involving persons with intellectual disabilities, there is a need for customized support [49]. Participatory design in games for health projects presents a range of methods and a variation in participation levels, where children sometimes have the role of ideators [50],

but more commonly of informants and/or testers [46,51,52]. Importantly, participatory design does not automatically translate into more effective games for health. One meta-analysis showed that involvement in some stages, such as in ideating a game's esthetics, may even be counterproductive and that children should preferably have roles as informants and co-creators of game challenges [43]. In addition, excessive participation might be energy draining for vulnerable participants and therefore ethically unwise [48]. Having a disability could thereby reduce the possible level of participation, if judged by personal presence. Total participation may thus be both unfeasible and unethical, and entail risks of decreasing the design quality. In order to address these issues, the design method *personas* can be implemented in design projects as a tool for maintaining a user perspective at stages where user engagement is problematic [48,53]. From a child perspective, the personas method offers a longer period of representation throughout the design process than if the child would represent himself/herself in person at only a few stages.

Personas

Personas is a critical user-centered methodology for orienting designers toward user goals by generating qualitative composite archetypes (user profiles) based mainly on qualitative user group data [54,55]. Data-driven personas help designers steer away from assumptions or stereotypes and instead focus on user preferences [56,57]. Furthermore, personas trigger empathy and new ways of thinking [57]. It is thus a goal-driven methodology, which fits with salutogenic approaches. The methodology originated in the interactive design domain, based on the reasoning that if you design for a specific user (visualized as a persona), you will be more successful in reaching users, than if you target "everyone" [54]. Following the introduction of personas in an increasing number of domains, it has consequently been used in the development of games for health that target children [9,41]. Personas move beyond statistical and demographic profiles as they include more detailed, rich, and engaging descriptions. Quotes, images, and details help shape a personality with an individual approach and life situation. Storytelling elements are common to flesh out personality and context (eg, a short story, a day in the life, a situation connected to a specific context, and a more general biography) [55,56].

In order to become believable characters that can have an impact on design, personas must be based on real data and created with consideration for the intended use [56,58,59]. The method requires merging qualitative (and sometimes quantitative) data from numerous people into convincing semifictional characters. Multiple personas can increase usability in the final design, but to rank and limit the number of personas are recommended to keep the design process manageable [55,59]. Personas can be validated through approval from the research team or potential stakeholders [60] and confirmation from participants [59], or through comparison with the data to ensure accurate reflection [55]. Given its flexible user-centered approach, personas as a method lends itself to participatory construction. However, this has not been extensively studied in the context of children with disabilities, as a general method for this is lacking. Against this background and with the described approaches, the personas methodology was seen as a suitable approach for further development through this study.

Objective

The objective of this study was to investigate how norm-creative and participatory design approaches can be integrated in a persona generation method to suit children with disabilities in the design of games for health. The aims of such a persona generation method would be easier involvement and stronger representation of children with disabilities in design processes where they are the target group.

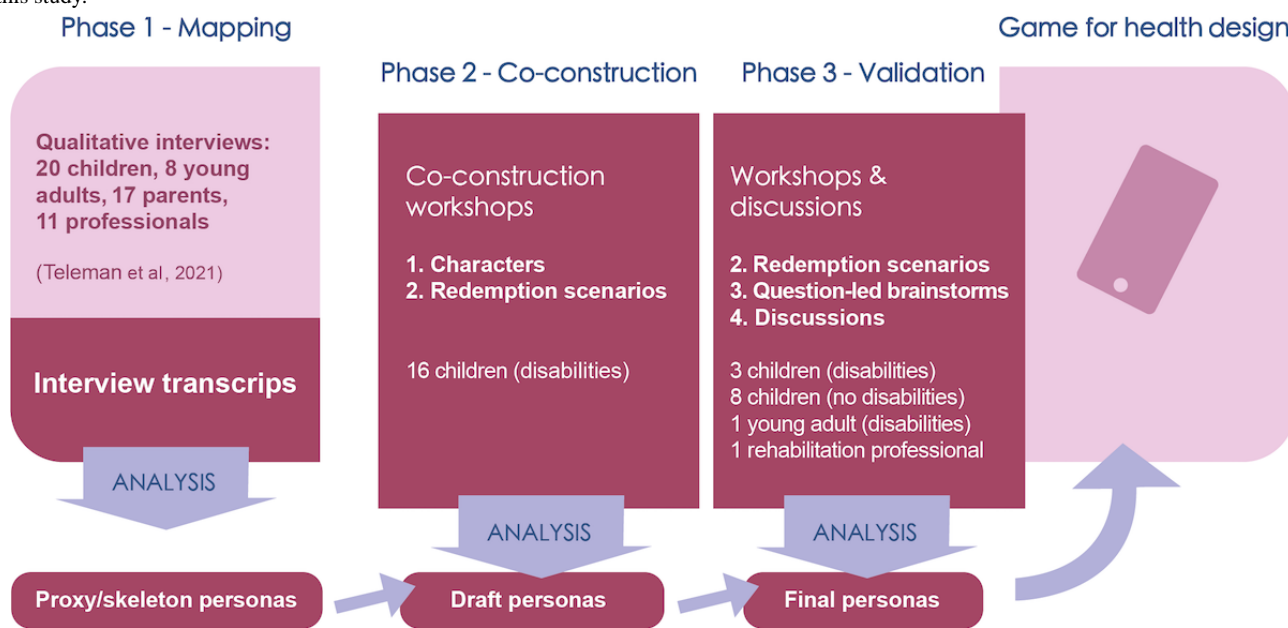
Methods

Setting and Study Design

Ethical approval was granted by the Regional Ethical Review Board at Lund University, Sweden (No: 2017/707).

In order to contextualize method development, a game for health design process conducted at a university college was used as an empirical case. The development was thus conducted so that the personas generated through this method could be applicable in the design of such a game (Figure 1). The target group for this game for health was children with disabilities, for whom it was to function as a digital decision support tool (accessed as a tablet/mobile app) to strengthen participation in decisions related to pediatric rehabilitation.

Figure 1. Process overview. The darker parts were included in this study (interview transcript analysis, co-construction, validation, and persona modeling) within the context of a game for health case. The application of the generated personas in the design of the game for health was not covered in this study.



A persona generation method was merged with a participatory design method, inspired by the work of Pruitt & Adlin [55] and Spinuzzi [34]. This merger was further remodeled to include steps where children could co-create characters and scenarios as proposed by Wärnestål et al [48]. The result was a unique study design with 3 main phases (Figure 1). It enabled interpretation and meaning creation from participant input in multiple iterative steps. The emerging data were explored through an inductive thematic design analysis. The 3 phases were as follows:

- (1) *Mapping phase.* Data (interview transcripts) from the overarching games for health case were utilized. It involved extracting and analyzing *factoids* (pieces of information) from the data to construct proxy/skeleton personas.
- (2) *Co-construction phase.* This phase consisted of creative workshops in which children co-constructed characters and redemption scenarios through images and storytelling. The output was analyzed to draft personas that were continually enriched and then ranked.
- (3) *Validation phase.* The draft personas were used in workshops and discussions with the target group and other actors, which contributed to the validation and finalization of the personas.

Recruitment and Participants

The interview transcripts analyzed in Phase 1 included the following 4 groups: children with disabilities, young adults with disabilities, parents of children with disabilities, and professionals working in pediatric rehabilitation (Figure 1). Only children were recruited for the workshops in Phases 2 and 3. The inclusion criteria were age 6 to 18 years and having an

established contact with pediatric rehabilitation services in southern Sweden, thereby having one or more disabilities. The children had to be able to participate in a workshop setting and communicate either orally in Swedish or via any of the augmentative and alternative communication tools used in their rehabilitation. These criteria were based on the resources and skills of the research team. Professionals helped determine which children met the criteria, and these children were invited to participate. The participating children were 6 to 17 years old and had a sociodemographic spread. There were 10 females and 6 males. Various disabilities were represented, including physical, cognitive, and intellectual disabilities, and autism spectrum disorder. Some children had multiple disabilities. Both the recruitment and workshops involved a speech and language therapist experienced in rehabilitation work with children with disabilities [53]. An additional group of children aged 10 to 12 years without disabilities was recruited through a local school for some validation workshops (question-led brainstorming). These workshops aimed to assess the personas’ usability in a design activity, which could involve people outside of the target group (see the Ethics section). The validation phase also involved 1 young adult with a disability (ie, formerly in the target group) and 1 professional working in pediatric rehabilitation.

Data Collection

An iterative persona modeling process took place during all study phases, informed by the analysis of each phase as shown in the bottom row of Figure 1. Each phase involved different activities, for example, rounds of workshops. A workshop overview is presented in Table 1.

Table 1. Workshops performed in Phases 2 and 3.

Workshop	Purpose	Participants	Activity	Output	Persona modeling
1. Characters	Get insights on the target group's <ul style="list-style-type: none"> • Life situation • Personality • Interests • Frustrations • Goals 	Children with disabilities	Co-creating collages of visual characters using templates and an image bank	Thirteen child characters	Drafts of 3 portrait-and-bullet point personas merged from characters and skeleton personas Simplified personas
2. Redemption scenarios	Get insights on the target group's strategies to manage frustrations Test and validate the personas' credibility and usability for the target group	Children with disabilities	Co-creating strategies in scenarios where personas encounter frustrating situations	Eight redemption scenarios	Persona stories (life situation) Nicknames (personality) Quotes (personality and/or life situation)
3. Question-led brainstorming	Test and validate the personas' credibility and usability in a design activity	Children without disabilities	Brainstorming on how a game for health could cater for the needs and interests of a persona	Audio recordings, sketches, and notes	Three persona versions for different uses Finalizing personas

Phase 1 (mapping) entailed an analysis of transcripts from 56 semistructured interviews that were part of the ongoing game for health case [5]. The interviews aimed to give an understanding of which experiences and perceptions of participation the children and other stakeholders had. Questions revolved around potential barriers and enabling factors for participation in rehabilitation.

Phase 2 (co-construction) involved 16 children in the roles of both informants and co-creators. This phase consisted of creative workshops. They were either individual, pair, or group workshops (3-5 participants). Each workshop lasted 60 to 120 minutes and was aimed to enable children to contribute with knowledge and creativity through visual input. This input consisted of fictional *characters* in Workshop 1 and of comic strips called *redemption scenarios* in Workshop 2 [48]. The workshops took place in accordance with the participants' preferences, either at their local rehabilitation center or in their own home, at a time chosen by the family. Children could choose to be accompanied by an adult, although most of the children wanted to participate alone. Well-matched participants were considered important for the group workshops as feeling comfortable is essential for child participation [3,5,61]. Some children knew each other already, which was considered an advantage when forming groups. One workshop included a school class and took place in a classroom during school hours without teachers being present.

Phase 3 (validation) included testing the personas in the construction of redemption scenarios in Workshop 2 (these workshops thus contributed to both co-construction and validation) and in brainstorming related to the game for health case in Workshop 3. The personas were also discussed with 1 young adult and 1 rehabilitation professional at the end of Phase 3. The brainstorm workshops were conducted in a classroom after school hours. There were 2 question-led sessions, with 4 participants in each. The participants were encouraged to write and draw their ideas, and the dialogue was recorded.

Data Analysis

Both data and method analyses were performed at the end of each workshop round. The analyses were first made individually (by BT and CK). They were then compared, merged, and discussed again in a larger group (including PS and JMN) [62,63]. A qualitative thematic design analysis inspired by Pruitt & Adlin [55] and Kolko [64] was used. Their analysis process is characterized by collaborative (1) visualization and mapping of insights/factoids (eg, by arranging sticky notes), (2) clustering and organizing, (3) finding and visualizing patterns, themes, and needs, and (4) summarizing [55,64]. A similar analysis process was used by Schulz & Fuglerud [53] to create adult personas with disabilities, and by Wärnestål et al [48] when co-creating child personas in vulnerable contexts.

Inputs from all participant groups were extracted from the interview transcripts and turned into factoids in the analysis of Phase 1. The factoids were then abstracted into themes describing user *needs*, which were clustered as notes. Transcripts were made of the visual data in Phases 2 to 3 (ie, output from the workshops). This included listing the images that were used in the co-created characters (and possible comments attached) to find emerging patterns. Visual data from the redemption scenarios were analyzed in a similar manner, where the children's stories were transcribed into factoids describing user *strategies*. The transcripts generated text or visuals that was mapped to different personas.

Ethics

Information about the study and the voluntary nature of participation was given to all the participants prior to inclusion. Informed written consent was obtained from parents (this term includes all legal guardians) for the children who chose to participate, as well as from participants over 15 years of age. All personal information was handled according to the General Data Protection Regulation [65] and the Swedish Ethical Review Act. Each child participated in a maximum of 2 participatory activities that were kept short in order not to drain the energy of participants, following the ethical principles of the World Medical Association [66]. While striving for user participation

at essential stages, some activities were considered justified to be performed with less vulnerable participants (children without disabilities/young adults with disabilities) for the same reason. Representation in these activities was based on the experience of being a child or having grown up with a disability [47,49].

Results

Phase 1–Mapping

Interview Transcript Analysis

We started to gain an understanding of the participants’ life situations and approaches, and could identify various *needs* in

the analysis of interview transcripts. The analysis showed that many of the children were not very talkative unless they had a chance to discuss their own interests, hobbies, or specific issues such as problems with their assistive technology, wheelchair, or similar. Many of them seemed to count on adults to communicate their needs and resources. The interview transcripts also showed that communication through speech was not always easy, and the total data from child interviews were limited in comparison with that from the other participant groups. However, many of the children considered themselves to be creative and good at problem solving. Figure 2 shows examples from the analysis of the interviews with the children.

Figure 2. Examples from the data analysis of transcripts from the interviews with children during the mapping phase.

Raw Data	Factoids	Themes (described as needs)	Persona input (addressing needs)
Transcript Coming up with ideas I am best at.	Good at coming up with ideas	Space and communication for own ideas	Quote: “I like challenges and solving problems”
What do you want to talk about in rehabilitation? Mm. [looking for a long time]. Oh. Can't find it? Nah	Hard to find words for thoughts	Help articulate thoughts and feelings	Frustrated when others don't understand me
Would you like to decide more? It's only the football I want to decide about.	Wish to discuss interests. Interests are important.	Motivation from personal interests	Quote: “Can we talk about football instead?”
Only the personality, if I get angry and stuff ... then let me be. So, in sick-, then, in the camp, I didn't think it was important for them to know about the dystrophy myotonic and ADD and ... and like ... and then everything else.	Feel safe when professionals know about my diagnoses	Provide and transfer information about myself	Characteristic: Intense, has a temper
Just like [last teacher] told [new teacher] what we normally do.	Good when professionals exchange routine information, so I don't have to	Provide and transfer information about myself	Don't like: Substitute teachers who don't know me

The interviews with the parents contained rich descriptions of what their children’s daily lives look like. Many expressed concerns regarding their children’s low level of participation and communication struggles. Rehabilitation professionals emphasized the importance of knowing and being “on the child’s level” to be able to communicate. Parents and professionals provided many examples of the children’s needs and how they personally work to accommodate these. Making rehabilitation exercises more playful and planning ahead were examples of this.

Modeling: Proxy/Skeleton Personas

A persona construction was initiated based on the mapping phase analysis, and 3 proxy/skeleton child personas were created (Multimedia Appendix 1). Their main purpose was to function as communication tools within the research team and with

potential stakeholders in the games for health case. The proxy/skeleton personas helped the team reach consensus about what they should contain and thus which ingredients to search for in order to create the final personas. They provided hints of what kinds of situations, goals, and issues children in the target group are dealing with. These initial versions thus also served as skeletons when developing the final personas.

Both our analysis and proxy/skeleton personas were disproportionately influenced by adults’ perspectives due to the uneven distribution of data. In order to address this, the following phases would have to enable children to express themselves through other means than interviews. Phase 2 therefore consisted of participatory workshops (1 and 2) with the goal of generating inputs that could enrich the proxy/skeleton personas (Table 1).

Phase 2—Co-construction

Workshop Preparations and Image Bank

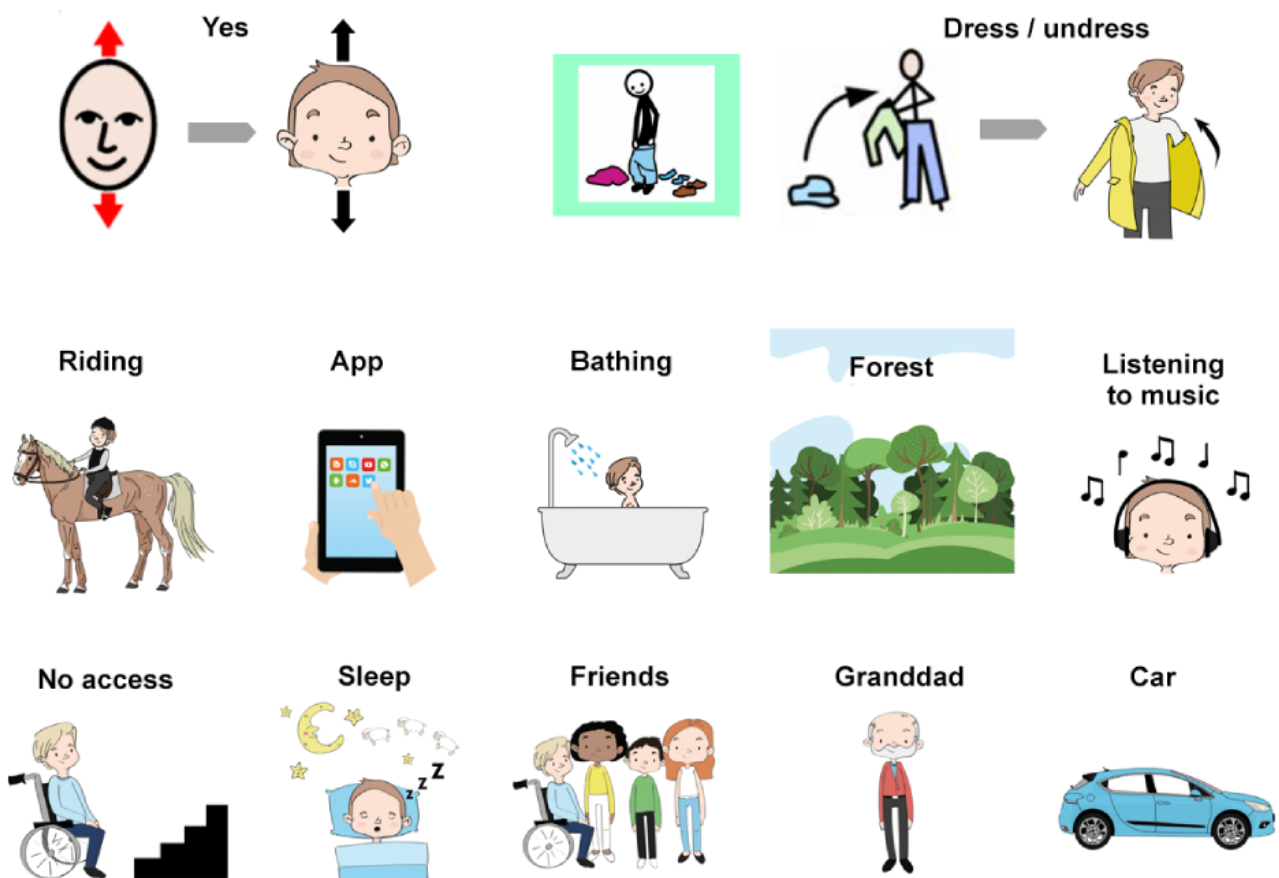
The purpose of Workshop 1 was for the children to generate fictive characters based on themselves. These characters were later to be merged, possibly with the proxy/skeleton personas, and remodeled into final child personas. A visual overview of the design process and a workshop agenda were presented (Multimedia Appendix 2) to gain trust and provide transparency. The workshop facilitators described the role of the children and emphasized the importance of their contribution as users of pediatric rehabilitation. It was also explained that there were no “solutions” or “right answers” to the activity. The facilitators wore casual colorful clothes and sat down with the children as opposed to standing above them to further mitigate the power imbalances between researchers/adults and participants/children. If there were fewer facilitators than children, they moved regularly between participants. It was preferred to not greatly outnumber the children as this could feel intimidating (maximum 2 facilitators per child).

The co-construction workshops had to offer other ways of participation than oral communication. The material also had to suit children of various ages and with various abilities and literacy levels. Visual and physical media allowed the participants to use nonliterate skills. An image bank with cards

and character templates was created, besides the visualizations of the process and the agenda. A test workshop carried out with the research team led to some adjustments. It also generated characters of each team member. These served as presentation tools for facilitators as well as examples when describing the activity (Multimedia Appendix 2).

An image bank of approximately 160 image cards was created with the purpose of enabling and materializing nonverbal and nonliteral input. Semantics and visual references would determine the cards’ ability to motivate and trigger ideas in the participants. We drew inspiration from the most established communication tools used in Swedish rehabilitation to increase clarity and familiarity in the images and kept crucial elements for easy identification. For example, arrows showing movement, a red flash symbolizing pain, symbols for *Yes/No*, and other abstract words were only slightly adjusted to visually match the image bank (Figure 3, top). The redesign of established images was partly a matter of visual coherence and partly about motivation through providing attractive and playful materials. Other images were designed from scratch based on the topics brought forward by children and parents in the interviews, such as loneliness, wheelchair access, or school situations. Finally, we added cards that would help generate foundations for personas, such as different hobbies, moods, or relationships (Figure 3).

Figure 3. Examples of redesigning existing communication aid images (top row) and new image cards (middle and bottom row).



The imagery for the workshops was designed to avoid prejudices around, for example, family forms or gender, which followed the norm-critical approach. There were, for example, cards representing a “nuclear family” as well as separate cards for mum, dad, brother, and sister. We also included characters with ambiguous gender and less gender coded outfits. However, the professionals advised us to keep the cards “simple, stereotypical, low in details, and sharp contrast.” In view of the slightly contrasting advice from Wilder [67] that states that children with disabilities are more likely to understand realistic images, we kept cards with animals and objects fairly realistic and less stylized or cartoon-like. It was, however, a question of balance between representation, comprehensibility, and practicality. A limitation on the variety of cards was that we had to keep the number of cards low in order to make the workshop manageable. Each representation of a person could not come in many variations of skin tones for example. The standard face created for the communication cards was slightly cartoonish and androgynous, with brown hair and a relatively fair skin tone (Figure 3, top left). The skin tone generally matched the children who participated in the workshops but could have been altered otherwise. Black and white images were not considered an option, as it would reduce both contrast and appeal. The workshop facilitators encouraged the participants to adjust the cards with the help of pens, glue, and scissors, as we had to use some archetypes where people, things, and places were concerned.

The image bank was to enable and motivate children to elaborate around both tangible and abstract concepts and generate cornerstones for the personas. It included sets with the following themes:

1. A template character, with a choice of accessories, costumes, and items to dress it with and color.
2. Communication aid cards such as *good, bad, approve of, now, not, thank you, who, know, and boring*. These were scarcely used since participants managed well without them.
3. Moods and personal characteristics such as *smart, clever, sad, crazy, nervous, curious, talkative, and fun*.
4. Actions such as *read, talk, ask, sleep, remember, teach, listen, look, give, and hurt*.
5. Chores such as *to clean, shower, dress, do homework, brush teeth, and go home*.
6. Hobbies and interests such as *bake, cycle, swim, read, music, sports, horse, and bird*.
7. Products, aids, and gadgets such as *wheelchair, tablet, TV, and image chart*.
8. People such as *mum, dad, grandparents, friends, teacher, assistant, and health care professional*.
9. Places such as *home, school, hospital, rehabilitation center, forest, and sea*.
10. Transports such as *car, bus, airplane, boat, and taxi*.
11. Struggles such as *crowds, no wheelchair access, difficulty to focus, pain, and vision problems*.
12. Blank cards for emerging ideas.

Workshop 1: Co-constructing Characters

The activity was briefly introduced and each child started with an A3 template with a blank character in the middle, items to dress it with, and piles of image cards on the side. The image cards were arranged in themes so that the participants could create collages around each theme.

It emerged that it was preferable for 1 adult to sit with each child, not too close to other participants, and guide the child through the process. Although the approach was to ask about the child’s own experience and life situation, a persona is not an actual person, which enabled some fantasy to go into the characters. The participants’ energy levels and attention span influenced the length of the workshops, which lasted between 60 and 120 minutes. This meant that participants considered themselves ready at various stages, which made us discard our initial idea of collectively summing up and reflecting on each character. Notes to remember or explain parts of the characters were added to the collages by the facilitators. The facilitators summarized their observations after each workshop and reflected on what could be improved for the following workshops. This could lead to complements or adjustments to the workshop layout or image bank. Each workshop contributed to a greater understanding of the participants’ life situation and task management.

Workshop 1: Output and Analysis

The output from each child (13 in total) was a colorful paper collage of image cards, drawings, and notes that together formed a part biographical part fictional character (Figure 4). The characters were transcribed into lists within the following 7 categories: personality, important relationships, hobbies/interests, frustrations, places, transports, and products, in order to visualize patterns in the data. Each image card’s frequency was also listed to provide insights on common topics within each category. Comments and expressions were also included. Observations of task management could also become persona input, such as whether a persona prefers process control or has a more discovery-oriented approach.

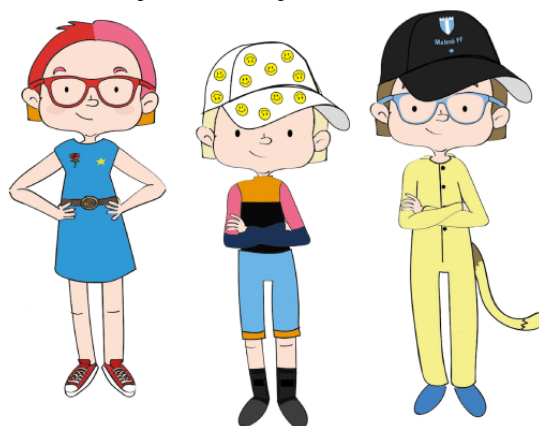
The analysis of the characters showed the use of similar cards for describing *personalities* and *frustrations*. Many characters displayed difficulties in expressing themselves or that others had difficulties in understanding them. One example was the statement “my parents need help in understanding me.” Figure 5 shows examples from the analysis of Workshop 1.

Observations of the workshop procedure showed that the image cards and layout had worked well in terms of allowing creativity. The children were imaginative and did not hesitate to adjust the image cards. Some also drew or wrote their own ideas. However, some parts did not suit the participants' fine motor skills. Small cutout costume pieces (eg, hats) proved difficult to deal with and had to be redesigned. A major part of the facilitation was to help participants stay focused or to move on to the next theme. It became clear that it was more difficult to concentrate in large groups than in individual or pair workshops. This was probably due to both noise and distraction levels, and that the facilitators had to walk around between participants. We reduced group sizes to have 1 facilitator per child further into Phase 2, which provided a calmer environment and more effective assistance. Some participants with a disability from the autism spectrum preferred a more structured process. We arranged the themes in order to address this and made an overview sheet showing all the cards within each theme. We also realized that if the participants became bored or tired, they might consider the workshop as finished. In order to help them distribute their energy evenly during the activity, we had to clarify what was expected to be covered on the A3 paper. Spending too much time creating the character in the middle (which could easily be perceived as the main part) could thereby be avoided.

Modeling: Creating Personas

In order to create personas out of the 13 co-constructed characters, they were divided into 5 groups based on similarities in *personalities* and *frustrations* (not disability). Three personas were considered suitable for this study. Three of the groups were thus selected, representing a variety of the aforementioned aspects. Each group was then merged into 1 persona by combining elements from all its characters. The personas now had a visual portrait and bullet points under the headings: I am good at, Family, Personality, Frustrations, Goals, Motivations, Products, I like, and I don't like. We prioritized input from children we knew wanted to contribute again when choosing between equivalent alternatives. Bringing forward visual details from the characters could generate sympathy from both recurring and new participants in the upcoming workshops. The proxy/skeleton personas were revisited and were used to flesh out the new personas. Adjustments were made to ensure that the personas expressed coherent characteristics and abilities. Personas were kept androgynous in terms of name and appearance when based on children from different genders. Appearance and clothing were influenced or copied from the children's characters. Assistive technology or aids were described, but disabilities were not explicit. [Figure 6](#) shows the visual portraits of the 3 personas *Molly*, *Kim*, and *Alex*.

Figure 6. Visual portraits of the personas, modeled from input of Workshop 1.




Workshop 2: Co-constructing Redemption Scenarios

The purpose of Workshop 2 was to flesh out the personas by gaining insights on the participants' problem-solving strategies in frustrating situations. Validation in these workshops (validation continued in Phase 3) entailed testing the credibility and usability of the personas. The participants co-created stories in the form of *redemption scenarios*, and we could see how the personas were perceived by the target group. The redemption scenarios were based on frustrations that had emerged in the characters from the previous workshops. Some contained more than one frustration. The first panel showed a situation, and the last panel showed that the situation was resolved and the persona was content. The children were asked to imagine how the story unfolds and connect the given beginning with the given end by filling out the empty panels in between. Six scenarios were designed, where 3 personas were represented in 2 scenarios each. Some related to rehabilitation situations but not all. It was considered more important to trigger strategic thinking in

general, than in connection to certain situations. Other people in the scenarios were open for interpretation (who they were), and the scenarios used generic words like "exercise" rather than specifying "weight training," "homework," etc. This was to enable many children to be able to relate. Different interpretations of contexts might also generate a variety of strategies. The activity in this workshop was more demanding than previous workshops since the scenarios involved another person (the persona) and problem-solving. The workshops were thus individual to offer more support. Simplified versions of the personas were made in order to make them easy to understand ([Figure 7](#)). These were presented prior to the scenarios. The children chose which scenarios to work with. They either drew and wrote themselves, together with facilitators, or they had the facilitator to draw and write while co-creating the narrative. The facilitators discussed the persona's character and interests, to trigger ideas in the participant. A little sketching by a facilitator could also stimulate the child's creativity.

Figure 7. Example of a simplified persona used for the redemption scenarios in Workshop 2.

Kim



Personality:
Fun & games
Loving
Clever

Likes:
Swimming
Horses
Kim's friends at rehab
Friends at swim school
Big sister

Things that can be tough:
Obstacles for wheelchair
Hard to sleep
Sometimes difficult to speak clearly

Good at:
Working out
Rolling my wheelchair
Horse riding
Walk with foot braces

Workshop 2: Output and Analysis



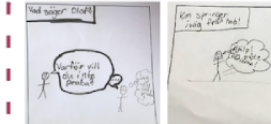
The output of Workshop 2 was 8 redemption scenarios (Figure 8). The events in the children's stories were abstracted into concepts of strategies (Figure 9). For example, people coming to rescue the persona were interpreted as *involve and getting*

help from people that you trust. Simplified solutions (lack of strategy) were found too. While some children spent a lot of time coloring details, others quickly moved on to the next scenario. It was evident that children who had participated in Workshop 1 appreciated seeing details from their previous creations.

Figure 8. Examples of co-constructed redemption scenarios in Workshop 2.



Figure 9. Examples from the data analysis of co-constructed redemption scenarios in Workshop 2.

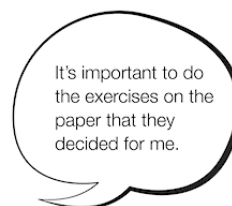
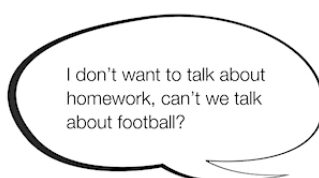
Raw Data	Factoids	Themes (described as needs)	Persona input (addressing needs)
<p>Visual</p> 	<p>Strategy: prove their case against too high demands</p>	<p>Communicating individual needs and resources</p>	<p>Frustration: Supplement teachers who don't know me</p>
	<p>Strategy: involve and get help from people you trust</p>	<p>Support from trusted persons</p>	<p>Quote: "My cousin is my idol and my assistant is a good friend of mine."</p>
	<p>Lacks strategy to communicate (avoid conversations)</p>	<p>Help to articulate thoughts and feelings</p>	<p>Dislikes to talk, prefer socialising online</p>

Modeling: Adjusting Personas

Based on the analysis of the redemption scenarios, the personas were slightly adjusted, and a story was created for each of them. In order to keep the personas open for different uses during the design process, the stories were not connected to a specific product or service. They were instead focused on the personas' life contexts and served as a descriptive complement to the existing bullet points. The 3 versions (bullet point, story, and simplified persona) could be used for communication with different participants and stakeholders depending on the activity. The personas were also given descriptive nicknames to quickly convey their character (eg, *Alex – the shy, organized expert* and

Molly – a cheerful, short-tempered leader). Each persona was completed with 2 quotes, such as "It's important to do the exercises on the paper, that they decided for me" and "I'm afraid I'll say something stupid when I'm angry and lose friends." The personas were then ranked, which resulted in 1 primary persona and 2 secondary personas. This prioritization was based on needs and with the game for health in mind. The primary persona (*Alex*) was considered to have most to gain from the game in question. Accommodating *Alex's* needs would probably also increase the appeal and usability for users in general. **Figure 10** shows the persona additions after Workshop 2 (quotes, nickname, and story).

Figure 10. Persona additions after Workshop 2 (quotes, nickname, and story).



Alex is a fairly quiet person, who thinks and reflects a lot. Sometimes thinking can turn into brooding, which consumes energy. Alex needs to fully understand things to be able to move on. Alex may be perceived as a little shy, because Alex often wants to think before talking, especially with new people. It can therefore take a while before the answer comes, and it can also be difficult to hear it clearly. It can be tricky to read Alex, which becomes an obstacle for new acquaintances. When tackling a new situation or problem, Alex starts from what feels right in the moment, without thinking so much about what others think. Alex has unwavering in opinions and takes for granted that others will understand. It can feel awkward to have to make a decision, if Alex is not usually allowed to do so in a situation. To come up with constructive solutions, it is good to get some help to understand what is required and why. Alex may just shut down if it feels too complex or uninteresting. Then Alex drifts away in thoughts instead.



Alex feels secure in routines and likes to keep track of the week's different activities and hours. Alex is orderly and thorough. It can be frustrating when things have to move fast, such as showering in the morning, and you do not have time to be careful. Then Alex prefers to shower in the evening instead. It is important for Alex to be able to take care of personal chores and approaching adulthood feels motivating. Alex's biggest motivation is football - watching matches, following the standings and knowing the names of different players and teams. Some products are also important to Alex, who always brings pencils so that drawings will always be good. Alex has plenty of energy when it comes to football, and has become something of an expert on the subject. Alex could easily talk football non-stop, so it can be difficult for those who want to talk about something else.

Phase 3–Validation

Workshop 3: Question-led Brainstorming

Validation of the personas started in Workshop 2 as described above and continued in Workshop 3 through question-led brainstorming with children without disabilities. The primary persona was tested as a brainstorm tool in this workshop to confirm its credibility and usability. The purpose of the activity was to gain input and feedback on design sketches of the game for health. The persona *Alex* was presented in the simplified version (Figure 7) as a target user for the game. Sketches of the game were presented, and discussions around its theme, logic, and visuals were initiated. *Alex* was used to engage the children in inquiries such as “If Alex feels worried, how can Alex convey this within the game?” or “If Alex struggles with keeping focus, how should questions and rewards appear?” The workshop output consisted of drawings, notes, and audio recordings. The persona was effective in that the participants were able to discuss issues from *Alex*’s perspective as well as from their own, expressing, for example, “Like, I’m thinking of Alex now, and football, referees, dunno... football players” (when discussing supportive sidekicks in the game) and “you shouldn’t focus on what you can’t do, but what you can do.” Representation was perceived as important. The participants initiated discussions about how the sketches reflected norms related to appearance and the importance of representative illustrations to enable children to identify with figures in a game.

Validation Discussions

The personas were discussed with 1 young adult with a disability and 1 pediatric rehabilitation professional at the end of Phase 3. While the brainstorm workshops tested the primary persona, these discussions covered all 3 personas. This led to minor rephrasings such as replacing “wheelchair dependence” with “uses wheelchair” to connote mobility rather than dependence. The overall perceptions of the 3 personas were summarized as follows:

*I absolutely think they feel credible. [Young adult]
[they] look good, many children/young people who are to be able to identify themselves and I think they’re comprehensive, which is positive!
[Rehabilitation professional]*

The final personas are presented in [Multimedia Appendix 3](#).

Discussion

Principal Findings

Persona Method Development

The method described involved development of existing persona generation and participatory design methods, adjusted to suit children with disabilities. The co-construction of characters and redemption scenarios offered a broad understanding of the participants’ life situation and approach, constituting a solid foundation for persona generation. Key factors were identified and addressed during the development. A substantial addition was the image bank, designed to motivate children and offer nonverbal construction of personas. The children were able to

cope with a large number of images as long as they were arranged in a manageable way, and a clear activity overview was provided. Another component was the simplified persona versions, made to be comprehensible to child participants in participatory activities. The study was based on visual elements and proposes a thematic analysis for abstracting and translating image-based data into personas. We found that small groups and individual support helped children to stay focused, which is in line with other participatory research with children with disabilities [44]. An iterative process with numerous steps and co-creators implicated a gradual validation of the generated personas and their usability.

The norm-critical and social model approach also distinguishes this persona generation from established methods. For instance, disability was not explicit in our personas, who instead displayed assistive aids, context-bound barriers, or *frustrations*. This deviates from, for example, the report of Schulz and Fuglerud [53], who suggested that personas should display both the disability and its effects on life. However, the social model perspective resonated with the data where barriers were sometimes described as external factors, such as a lack of wheelchair access, or in quotes like “my parents need help in understanding me.” This contrasts pathogenic perspectives and norms regarding dysfunctions, their origins, and who has a problem. Similarly, since both gender and disability were regarded as partially socially constructed, some of the personas had no explicit gender. The fact that the target group was so diverse was an additional reason for keeping some characteristics open for interpretation and identification. Another deviant detail in our method was to use illustrations instead of photographs [56] to visually portray the personas. This was considered playful and enabled us to easily use elements from the children’s characters.

Finding the Right Level of Participation

If we acknowledge that people affected by design and technology should participate in its development, it is important to consider power imbalances and who has authority to select or neglect knowledge [12,68-70]. Although this study emphasizes increased participation for children with disabilities, it might not satisfy demands within participatory research for participant involvement through *all* stages, including the analysis [34,68]. Skills and capabilities are often carefully considered for this group to determine suitable participation levels for each participant [11,43,44]. Caution in terms of extensive activities for vulnerable participants has also been discussed in this paper, as children were not involved in all the analysis and validation steps. Instead, numerous participants and iterations of activities meant a continuous adjustment and a gradual validation of the analytical output. Furthermore, since disability is just one of many variables for our target group, they share many preferences with children without disabilities. Users outside of the target group were thus considered as valid representatives for some of the activities [47,49].

Recruitment and Representation

The difficulty in accessing and recruiting child patients and participants with disabilities has been recognized in previous research [9,46,49]. Both parental consent and other forms of

engagement were necessary in this study, such as providing transportation to an activity. This can constitute a risk that the participants only represent groups where parents have an interest in increased child participation as well as have the resources to allow them to take part. Children's voices in research are normally conditioned by parental consent, which poses a dilemma since it narrows the spread of perspectives being heard [71]. One way to reach participants with diverse backgrounds in this study was to conduct some workshops at schools. It has been suggested that being situated within a school environment could hinder full participation due to lingering power hierarchies in the buildings [47] and that children might only do the minimum required [43]. Although this was not perceived to be a problem for the activity, it is difficult to rule out that motivation was lower in the school workshops.

Norm-Creative Tradeoffs

Bearing in mind that materials used in design processes inevitably shape their result [72], we paid attention to how the imagery portrayed family setup, gender, ethnicity, and functionality to avoid producing norm-affirmative output or stereotypical narratives. It was sometimes a question of balancing representation on the one hand, and comprehensibility or manageability on the other. We thus had to use some archetypes and a limited variety in the image bank. This limitation could be partly addressed by being attentive to the participants' preferences and encouraging them to add to the material. However, should this method be transferred to more diverse contexts, we suggest a more varied representation, for example, in the skin tone of characters. The designed material is thus not fixed but highly malleable to enable flexible applications.

Implications

Norm-criticism raises awareness of norms that exclude or discriminate, while *norm-creativity* is a combined approach requiring both norm-critical awareness and design thinking, with the aim to move beyond or counteract norms [39]. As norm-criticism influenced this study's creative process, the resulting method has potential to shape norm-creative solutions

in projects using it. However, norm-critical awareness must accompany its use as the method does not replace critical thinking in the researcher. As highlighted by Pruitt and Grudin [56], personas must be used ethically, as with all scientific methods. Personas are tools for both design thinking and norm challenging, which may occur during persona construction or in the use of personas. Discussions triggered by a norm-creative persona generation method can also generate reflexivity within research [71,73]. The described method enables increased influence for children with disabilities in research and design processes. This might in the long run also influence norms of decision-making within such contexts.

The method was developed to suit children aged 6 to 17 years with disabilities, but while the method might be transferable to similar groups, age might not solely dictate who finds it beneficial. Personality, physical abilities, or cognitive development could be equally important [12]. The facilitators' resources and communicative skills could also affect the possibilities for participation. While the results were influenced by a game for health context, the method can be adjusted to suit other design contexts too. There are unlimited possibilities in terms of materials that can help include participants. One limitation of this study was that no alternative materials or settings were compared. Future research could thus involve other contexts or user groups, as well as comparisons and usage of differently generated personas.

Conclusions

This paper describes the development of a participatory persona generation method aimed to suit children with disabilities. The method strives to enable and capture the perspectives of this group by using iterative workshops and flexible materials. The results provide guiding examples for image-based workshops and analysis. Combined with norm-critical awareness, the method has potential to influence design projects in the direction of increased representation, norm-creativity, and inclusiveness. The method was developed within a games for health case, through which it was contextualized and validated. It may also be suited for, or adjusted to, similar contexts or user groups. This could be subject to further research.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Proxy/skeleton personas.

[[PDF File \(Adobe PDF File\), 375 KB - jopm_v14i1e29743_app1.pdf](#)]

Multimedia Appendix 2

Project overview and workshop agenda.

[PDF File (Adobe PDF File), 161 KB - [jopm_v14i1e29743_app2.pdf](#)]

Multimedia Appendix 3

Final personas.

[PDF File (Adobe PDF File), 1140 KB - [jopm_v14i1e29743_app3.pdf](#)]

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Original Paper

Co-design of an Electronic Dashboard to Support the Coproduction of Care in Pediatric Rheumatic Disease: Human-Centered Design and Usability Testing

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Abstract

Background: The coproduction of care involves patients and families partnering with their clinicians and care teams, with the premise that each brings their own perspective, knowledge, and expertise, as well as their own values, goals, and preferences, to the partnership. Dashboards can display meaningful patient and clinical data to assess how a patient is doing and inform shared decision-making. Increasing communication between patients and care teams is particularly important for children with chronic conditions. Juvenile idiopathic arthritis (JIA), the most common chronic pediatric rheumatic condition, is associated with increased pain, decreased function, and decreased quality of life.

Objective: The aim of this study is to design a dashboard prototype for use in coproducing care in patients with JIA. We evaluated the use and needs of end users, obtained a consensus on the necessary dashboard data elements, and constructed display prototypes to inform meaningful discussions for coproduction.

Methods: A human-centered design approach involving parents, patients, clinicians, and care team members was used to develop a dashboard to support the coproduction of care in 4 ambulatory pediatric rheumatology clinics. We engaged a multidisciplinary team (n=18) of patients, parents, clinicians, nurses, and staff during an in-person kick-off meeting followed by biweekly meetings. We also leveraged advisory panels. Teams mapped workflows and patient journeys, created personas, and developed dashboard sketches. The final dashboard components were determined via Delphi consensus voting. Low-tech dashboard testing was completed during clinic visits, and visual display prototypes were iterated by using the Plan-Do-Study-Act methodology. Patients and clinicians were surveyed regarding their experiences.

Results: Teams achieved consensus on what data mattered most at the point of care to support patients with JIA, families, and clinicians collaborating to make the best possible health care decisions. Notable themes included the right data in the right place at the right time, data in once for multiple purposes, patient and family self-management components, and the opportunity for education and increased transparency. A final set of 11 dashboard data elements was identified, including patient-reported

outcomes, clinical data, and medications. Important design considerations featured the incorporation of real-time data, clearly labeled graphs, and vertical orientation to facilitate review and discussion. Prototype paper-testing with 36 patients and families yielded positive feedback, with 89% (8/9) to 100% (9/9) of parents (n=9) and 80% (8/10) to 90% (9/10) of clinicians (n=10) strongly agreeing or agreeing that the dashboard was useful during clinic discussions, helped to talk about what mattered most, and informed health care decision-making.

Conclusions: We developed a dashboard prototype that displays patient-reported and clinical data over time, along with medications that can be used during a clinic visit to support meaningful conversations and shared decision-making among patients with JIA, their families, and their clinicians and care teams.

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KEYWORDS

human-centered design; coproduction; dashboard; pediatric rheumatology; juvenile idiopathic arthritis; JIA; juvenile arthritis; patient centered; patient-reported outcomes; patient communication; patient education; family education

Introduction

Background

The coproduction of care involves patients and families partnering with their clinicians and care teams, with the premise that each brings their own perspective, knowledge, and expertise, as well as their own values, goals, and preferences, to the partnership. Inviting and integrating these unique strengths support effective patient-family-clinician relationships [1-3]. Recent studies have shown that these partnerships can also be aided by implementing dashboards that display meaningful data that can be reviewed together at the point of care to assess a patient's progress and to make shared treatment decisions, particularly for patients and families living with chronic illnesses [4-6].

Juvenile idiopathic arthritis (JIA) is the most common chronic rheumatic condition, affecting 1 in 1000 [7,8] children. Even with advances in treatment options such as biological medications, children with JIA have decreased physical function, worse health-related quality of life, and increased pain despite improved disease activity [9-11]. Although the pediatric rheumatology field routinely collects patient-reported outcomes (PROs) of pain, function, and disease activity for research and collaborative improvement purposes [12,13], these data are not regularly integrated into clinical practice to inform care and treatment decisions. Growing evidence suggests that leveraging such data at the point of care can lead to improved health outcomes, which are of critical importance to children and families living with JIA. These families manage complex treatment regimens and regular visits with multiple subspecialists, including ophthalmology, psychology, and physical and occupational therapists, and usually require the long-term use of injectable and infusion therapies [14]. It is also known that children, parents, and clinicians have different views of disease and expectations of treatment outcomes, and it is important to enable children to actively communicate their views with their clinician [15].

Dashboard data visualization tools are used in health care to aggregate and integrate key data for review and discussion during clinical encounters to support patient-centered care. The

Swedish Rheumatology Quality Register dashboard serves as a long-standing rheumatology model [16]. It integrates and displays PROs (eg, pain, global health, and fatigue), key clinical data (eg, joint count and disease activity scores), and treatments and medications longitudinally and has been used by patients and their clinicians since 2004 to engage in coproduction of care. The Swedish Rheumatology Quality approach was associated with a 50% decrease in disease activity between 2004 and 2014 in people living in Sweden with rheumatoid arthritis (RA) [17].

Dashboard use in the US rheumatology community is increasing, building on earlier work in the field to provide data for clinical decision support at the point of care [18]. Design efforts have been completed at the University of California, San Francisco, health system to support an electronic health record (EHR)-based patient-facing dashboard for adult patients with RA [19,20]. However, we are not aware of similar efforts in patients with pediatric rheumatology and their families. Given the dearth of evidence-based care protocols in pediatric rheumatology, these patients and their families face an even greater need to bring together PROs and key clinical data in one place to support shared decision-making. To address this gap, we developed a human-centered co-design process to create a prototype of an electronic JIA dashboard.

Objective

Our objective was to design a real-time point-of-care dashboard to support partnerships between patients and families and their clinicians by identifying the data and information that matter most to them and designing the display for enhanced communication and decision-making.

Methods

Overview

Our study was guided by a human-centered design process [21] to ensure that the final dashboard design would serve the needs and goals of end users. The process involved deploying a series of iterative methods to (1) explore the context of use and needs of end users and (2) achieve consensus on the dashboard data elements and overall dashboard design (Figure 1).

Journey Mapping

The parent partner advisory group participated in a journey mapping exercise [21] intended to capture the patient and family lived experience in the care journey. Journey mapping described “walking through a visit”—from preparing for the visit at home, arriving at the clinic, moving through the clinic visit, and checking out to following up afterward. Parent partners described actions, questions that needed answering, happy moments (things that improved the care experience), and pain points (frustrations and annoyances) for each step in the care process. The aim was to consider how to leverage happy moments and understand how to improve pain points in designing the dashboard innovation.

Dashboard Sketches

Teams engaged in a visual thinking exercise designed to generate ideas for dashboard design and invite commentary [24]. Each team member was instructed to draw a picture of their ideal dashboard, review and discuss it with their team, and compare and consolidate the best ideas to create a team dashboard. Similarities and differences across team sketches were noted and discussed during the co-design session.

Site Visits

The Dartmouth Institute for Health Policy and Clinical Practice team conducted a site visit at each pilot site to engage key stakeholders (team members, local leadership and informatics teams, and patients and families) in the dashboard design process and to observe clinical operations and care flows to better understand the context and workflow of the dashboard. They met with patients and families (n=12) in individual and group settings to provide an overview of the dashboard initiative and capture their feedback and ideas for enhancing the information environment at the point of care.

Building Consensus

After exploring how a dashboard could support the needs and requirements of patients, families, clinicians, and care teams for use during clinic visits, teams engaged in determining the detailed design requirements for the dashboard. This process included finalizing the data elements, layout, and visual look and feel of the dashboard, and considering features to support self-management and other user needs.

Delphi Voting on Dashboard Data Elements

Clinical care team members, patients, and parents participated in a Delphi voting process via web-based surveys and multisite team meetings to achieve consensus on a parsimonious set of data elements to be displayed on the dashboard. An initial list of data elements and dashboard features was compiled based on personas, dashboard sketches, and discussions with patients, families, clinicians, and care teams during pilot site design sessions and site visits. Teams were also asked to review the list and suggest additional items that might be missing.

Dashboard elements were organized by domain in preparation for the 3 rounds of Delphi voting. The domains included PROs, clinical data, medications, self-management, and other user features. In the first 2 rounds, team members ranked the elements within each domain using a 5-point Likert scale. They

also indicated their top 5 elements for aggregate reviews as a cross-check against the rankings. After the second round of voting, the domain containing self-management and other user features was removed from the final round of voting, as it was determined that the patient and family-facing self-management tool was outside the initial scope of the point-of-care dashboard.

In the third and final round of voting, team members were asked to prioritize the data elements based on modified MoSCoW (must have, should have, could have, will not have) criteria [25]: must have, nice to have, will not have (but nice someday), and not a priority. After the results from the third round of voting were tallied, a broader group of stakeholders—clinician advisory group, parent partner advisory group, and CARRA and PR-COIN registry leaders—were engaged in reviewing and offering feedback before the dashboard data elements were finalized.

Specific measures or tools for each PRO data element were reconciled and aligned with the data collection for the CARRA [12] and PR-COIN [26] registries, with the goal of data collected once and used for multiple purposes. In addition, measures or tools validated for use in pediatric rheumatology were identified as preferable.

Low-Tech Dashboard Testing and Design Iteration

Sites engaged in testing a paper-based version of the dashboard with a small number of clinical patients and families using the Plan-Do-Study-Act framework [27]. The aim of rapid-cycle testing was to assess the feasibility and utility of the dashboard at the point of care and to incorporate feedback to iteratively refine and enhance the dashboard design and usability within the flow of care.

The dashboard prototype was created using a Microsoft Excel [28] template to display patient-reported data and key clinical data obtained from patients completing previsit questionnaires and the EHR. It was introduced to patients with JIA and their families by a rheumatology clinician. Parents were surveyed after the visit to rate their overall impressions of the prototype dashboard. Clinicians were also surveyed regarding their experiences with the dashboard.

Following low-tech testing, a third-party digital health solution organization was engaged to translate the findings from the human-centered design process into a final wireframe of the dashboard design. Iterative design sessions were held to obtain feedback based on the preferences of the patients, families, clinicians, and care teams.

Results

Insights From Contexts and End-User Needs

Human-centered design activities demonstrated the needs of end users and the processes required to integrate a dashboard into the flow of clinical care. This iterative framework generated ideas and insights about the features and functionalities that are most important to the design of the dashboard. Several notable themes emerged from iterative discussions between the project facilitators and co-design teams throughout the co-design process: (1) the right data, in the right place, at the right time,

(2) data in once for multiple purposes, (3) patient and family self-management components, and (4) opportunity for education and increased transparency.

The Right Data, in the Right Place, at the Right Time

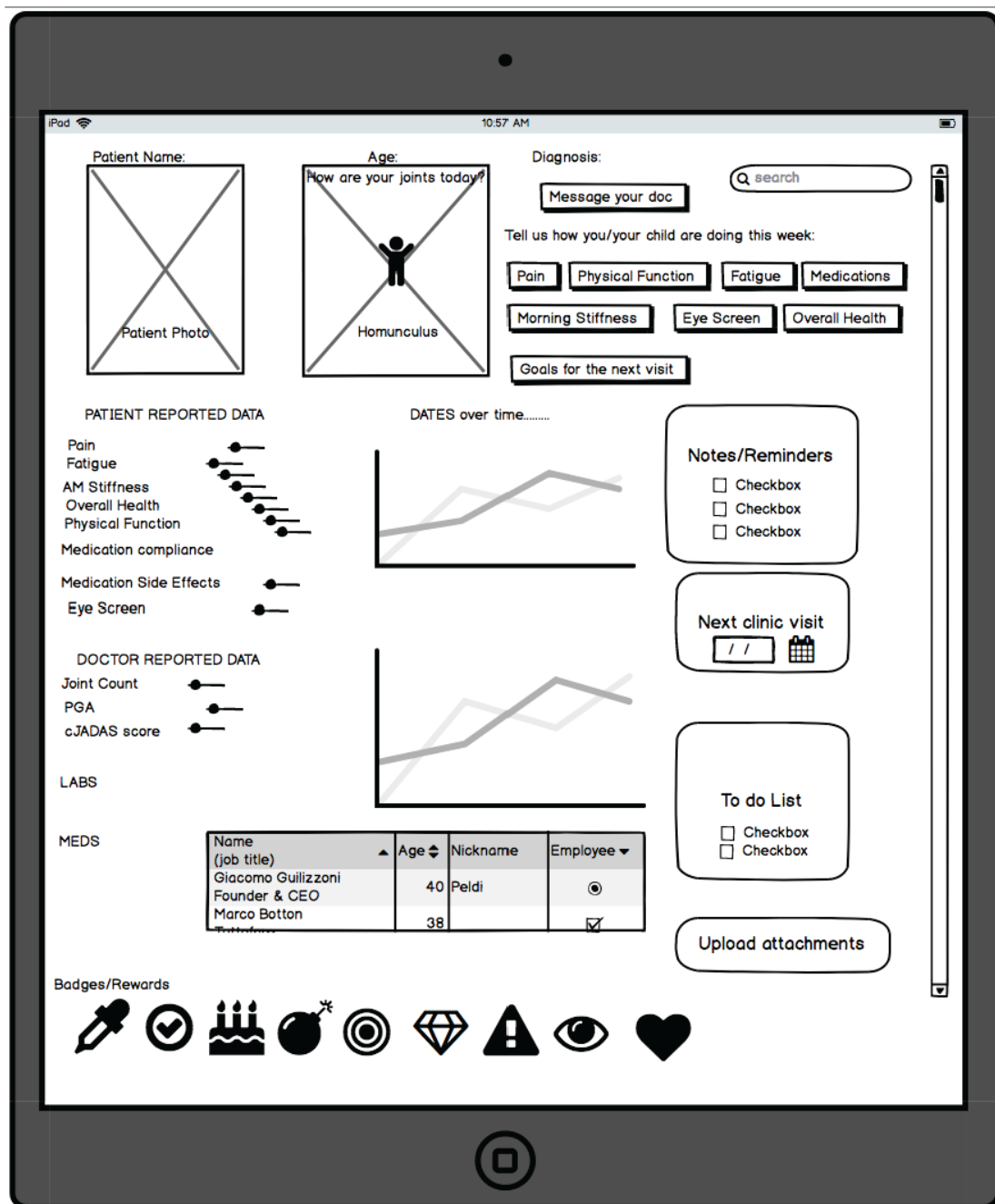
The EHR serves as a cumulative repository of the data and information generated at each clinical encounter. Pilot site teams stressed they did not want to replicate the EHR and instead arrived at a balance of data to support the coproduction of care:

Making decisions about treatment plans and medications is complex and involves weighing pros/cons...need enough/sufficient information but not too overwhelming. [Clinician: multisite team meeting]

Trending data over time (including medication usage) was identified as an essential functionality and included in all team dashboard sketches (Figure 2):

I would like to know how she is progressing better or worse over time. [Parent persona]

Figure 2. Example dashboard sketch. cJADAS: clinical juvenile arthritis disease activity score; PGA: physician global assessment.



In addition, the teams expressed a desire to personalize the dashboard with a patient photo and updates on life between visits:

We normally just see snapshots of patients in clinics; it would be great to have a bigger picture of what goes on in our patients' lives on a day-to-day basis.

[Clinician: multisite team meeting]

Data in Once for Multiple Purposes

A frustration identified by patients and families during the journey mapping exercise was being asked the same questions repeatedly during a visit as well as completing questionnaires and then not seeing the results or understanding how the data were being used:

A goal is to have information that gets shared actually get to the provider—by the time we get to the clinician, only 10% of what we've shared at every step of the visit process actually gets to the doctor.

[Parent: 2-day kick-off meeting]

In addition, clinicians discovered that the collection of PROs typically occurred toward the end of the visit, with a research coordinator collecting the data for research registry purposes. Teams agreed that an important design specification would be to ensure that previsit questionnaire data both inform the clinic visit and populate registries.

Patient and Family Self-management Components

Personas developed for this project, such as the example in [Figure 3](#), provide insight into patient and family needs in managing chronic diseases. Families of children and teenagers with JIA desire a place to collect, track, and review disparate pieces of information and data needed to optimize the management of their child's health:

I am looking forward to one place where my son's health information is all in one place for me to see.

[Parent persona]


A space for children and teenagers with JIA to self-report on activities or symptoms important to them, a *to-do* list, and a medication tracker were cited as desirable self-management features:

I would like a way to keep track of how I feel mentally and physically in between visits so that I can let my doctor know, especially since my visits are spread out.

[Patient persona]

Furthermore, during the co-design sessions, parents shared the information that they routinely collected to prepare for a clinic visit, including laboratory work required and completed, questions and updates to share with the physician, and the date of their child's last eye examination. They expressed the need to have this information centrally available for previsit planning.

Figure 3. Parent persona.

 <p>QUOTE: Sometimes a positive thought can help you through troubled moments.</p> <p>NAME: Vicki Senetti</p> <p>AGE: 40</p> <p>PROFESSION: Account Director</p> <p>BIO: Vicki is an Account Director for a communications company. Her daughter Sophie has Systemic Juvenile Idiopathic Arthritis (SJIA). Vicki is the primary caregiver and is responsible for ensuring Sophie takes all her medicine, attends medical visits and infusions.</p>	<p>INTERESTS:</p> <ul style="list-style-type: none"> • Party-planning • Spending time with family and friends • Netflix binge-watching • Dancing • Going for walks • Massages 	<p>SKILLS:</p> <ul style="list-style-type: none"> • Project Management and communication • Meeting and event planning • Leading project teams • Creating standard operating procedures • iPhone, iPad, laptop
	<p>GOALS:</p> <ul style="list-style-type: none"> • Continue to advocate for Sophie until she achieves remission • Ensure Sophie continues to be able to participate in all activities she is interested in • Prepare Sophie to be more independent as it relates to her health care as college is only 4 years away 	<p>DAILY ROUTINE:</p> <ul style="list-style-type: none"> • Prepare children for school • Work full-time • Drop off and pick up from Sophie's job or other activities • Cook dinner • Ensure Sophie takes her folic acid, Vitamin D and Methotrexate weekly • Take a walk when work is not busy • Binge watch shows on Netflix if I can stay awake
	<p>LIKES/DISLIKES:</p> <p>Likes</p> <ul style="list-style-type: none"> • Interactions with care team including rheumatologist, pulmonologist, nurses, infusion center staff, child life, etc. <p>Dislikes</p> <ul style="list-style-type: none"> • Waiting for medicine at times in infusion center • Lack of proactiveness to extend time between infusions 	<p>MOTIVATION:</p> <ul style="list-style-type: none"> • The idea of Sophie outgrowing this disease • Continuing to do what we are doing so Sophie can continue to do well
<p>CONTEXT: Vicki is Sophie's primary care giver – she is at every doctor visit, is the one ensuring she is taking her medication, is monitoring how she is feeling both physically and mentally, communicates with her doctors, nurses, school, etc. Vicki also works full time and has to be able to multi-task on a daily basis. The fact that information about Sophie's health could be at her fingertip is something that she is looking forward to seeing and that she thinks will be so valuable. She tries her best to keep track of everything but with such a busy schedule a tool that could help keep her organized would be extremely helpful and appreciated. Moreover, she is excited to be able to be connected with other caregivers, especially those that may be experiencing a similar journey in case the question of "what's next?" should come up.</p>		
<p>NEEDS/DESIRES: I would like a like a way to have Sophie keep track of how she feels mentally and physically in between visits so she doesn't forget anything worth mentioning in between visits. I would like to know how she is progressing better or worse over time. I would like to communicate with other parents that are going through the same journey.</p>		

Opportunity for Patient Education and Increased Transparency

Involving patients and parents alongside clinicians and care teams during the design process offered each group unique insight into the data and information most important to each in coproducing care. For example, parents were particularly concerned about medication side effects and lobbied to include a laboratory measure of liver toxicity (alanine aminotransferase)

on the dashboard. Clinicians and care team members assured parents that they always reviewed alanine aminotransferase scores as part of every visit; however, they deferred to parent preference to include it on the dashboard. Similarly, patients and parents were unaware of measures used for clinical assessment, such as the Juvenile Arthritis Disease Activity Score (JADAS) [29]. Clinicians admitted that they did not typically explain the clinical and research importance of the JADAS, acknowledging that the dashboard would offer an opportunity

for education and increased transparency with patients and families:

We would like to spend less time charting and dealing with insurance companies and more time with our patients on education and management of the disease, ensuring that we/they have a true understanding of their medical condition and treatments, daily life, and coping strategies. We want to promote self-reliance and self-management. [Clinician persona]

Consensus on Dashboard Design

Finalized Set of Data Elements

The Delphi method was used to reach consensus on the final set of dashboard data elements. The necessary data domains

Textbox 1. Final dashboard data elements.

<p>Patient-reported outcomes</p> <ul style="list-style-type: none"> Concerns, questions: free-text patient, parent, and family questions or concerns for discussion in the visit Patient global assessment: ordinal 0-10 scale, patient's assessment of overall well-being Physical function: Patient Reported Outcomes Measurement Information System (PROMIS) v1.0 Pediatric Upper Extremity short form raw summed score; PROMIS v1.0 Pediatric Mobility short form raw summed score Pain, pain interference: ordinal 0-10 current rating of pain and PROMIS v1.0 Pain Interference Short Form raw summed score Medication adherence: 5-point Likert scale that indicates how often medications are being taken as prescribed and includes the option of "I am not currently taking any prescribed rheumatology medications." Medication side effects: list of symptoms experienced with current medications <p>Clinical data</p> <ul style="list-style-type: none"> Joint count: total number of tender joints and total number of swollen joints Provider global assessment: ordinal 0-10 scale (provider's assessment of patient's overall disease activity) Disease activity (Juvenile Arthritis Disease Activity Score): 0-30 composite score that incorporates patient global assessment, provider global assessment, and joint count Liver toxicity: alanine aminotransferase <p>Medications</p> <ul style="list-style-type: none"> Medications: medication name, dose, route, start and stop dates, and frequency

Low-Tech Testing and Design Iteration

The paper-based prototype (Figure 4) was tested with 36 patients with JIA (aged 3-20 years; 24/36, 67% female) during a clinical visit. Parent (n=9) and clinician (n=10) feedback was very

included PROs, key clinical data, and current and past medications. The initial round of voting started with 25 data elements: 13 in the PRO domain, 11 in the clinical data domain, and 1 associated with medications. After 3 rounds of voting, a consensus was reached on the 11 data elements summarized in Textbox 1.

Data elements that were initially considered, but did not achieve consensus, included fatigue, morning stiffness, mental health, uveitis status, and several laboratory values (C-reactive protein, erythrocyte sedimentation rate, and aspartate aminotransferase).

positive (Figures 5 and 6), with 89% (8/9) to 100% (9/9) of parents and 80% (8/10) to 90% (9/10) of clinicians strongly agreeing or agreeing that the dashboard (1) was useful during clinical discussions, (2) helped to talk about what mattered most, and (3) helped to make health care decisions.

Figure 5. Dashboard low-tech testing: parent feedback (n=9).

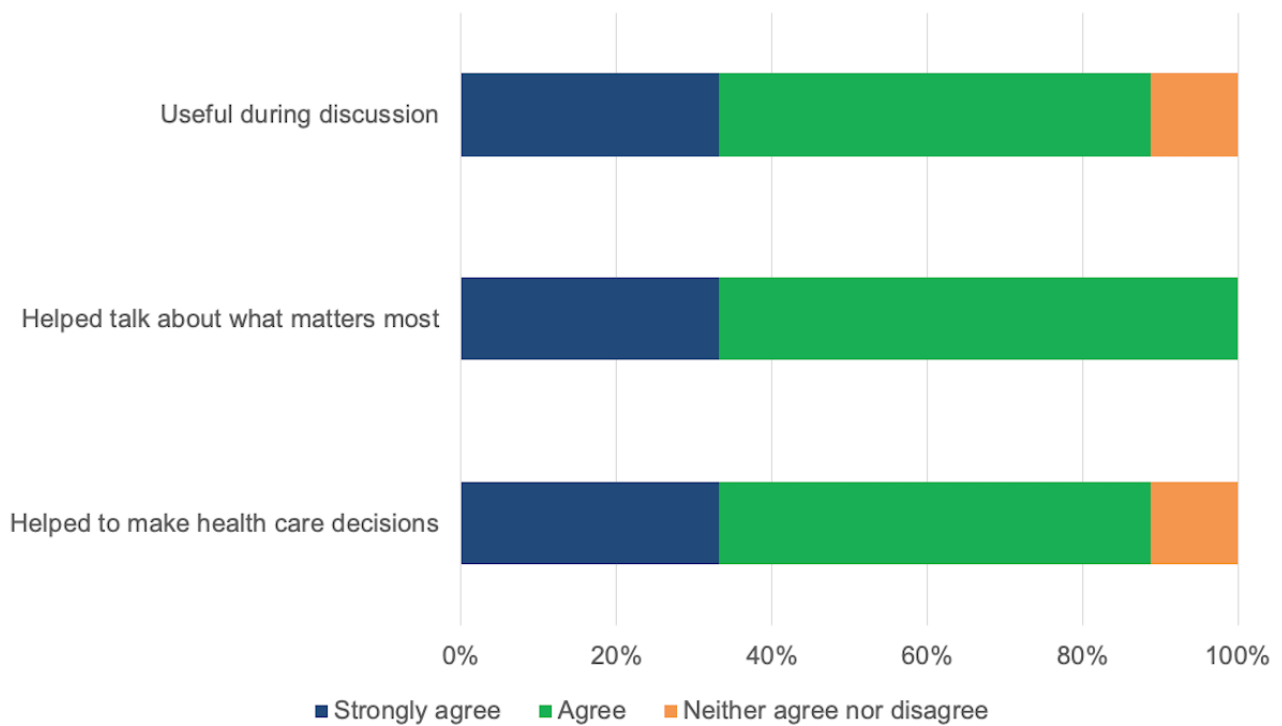
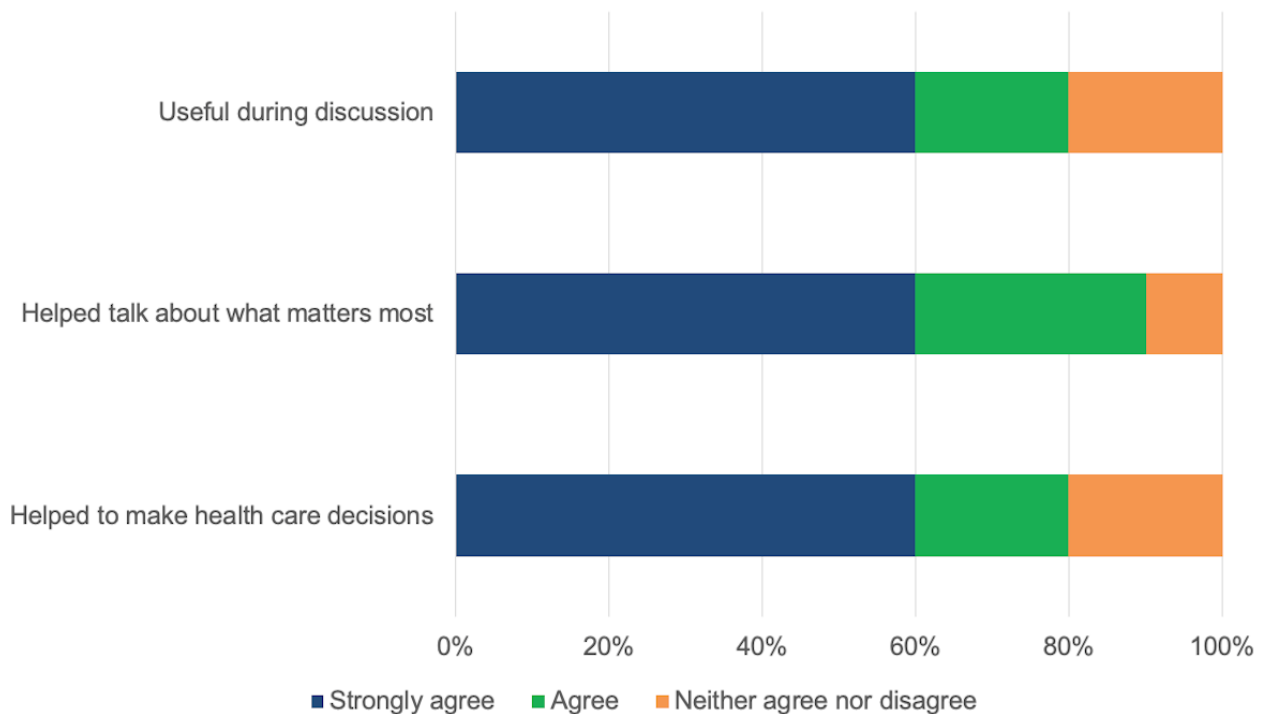


Figure 6. Dashboard low-tech testing: clinician feedback (n=10).



Low-tech testing also uncovered important insights about the value of the dashboard, including the ability of patients and parents to share questions and concerns in advance of the visit, greater transparency in clinical assessment data routinely collected by clinicians and care teams (eg, joint count, physician

global, and disease activity score), and the visualization of data over time to help make decisions. Although many PRO measures are still being validated for clinical use [30], patients and families are interested in seeing their scores in real time [31] and comparing them with past visits. The dashboard designs

were iterated to include these elements and to improve the visual interface throughout the study.

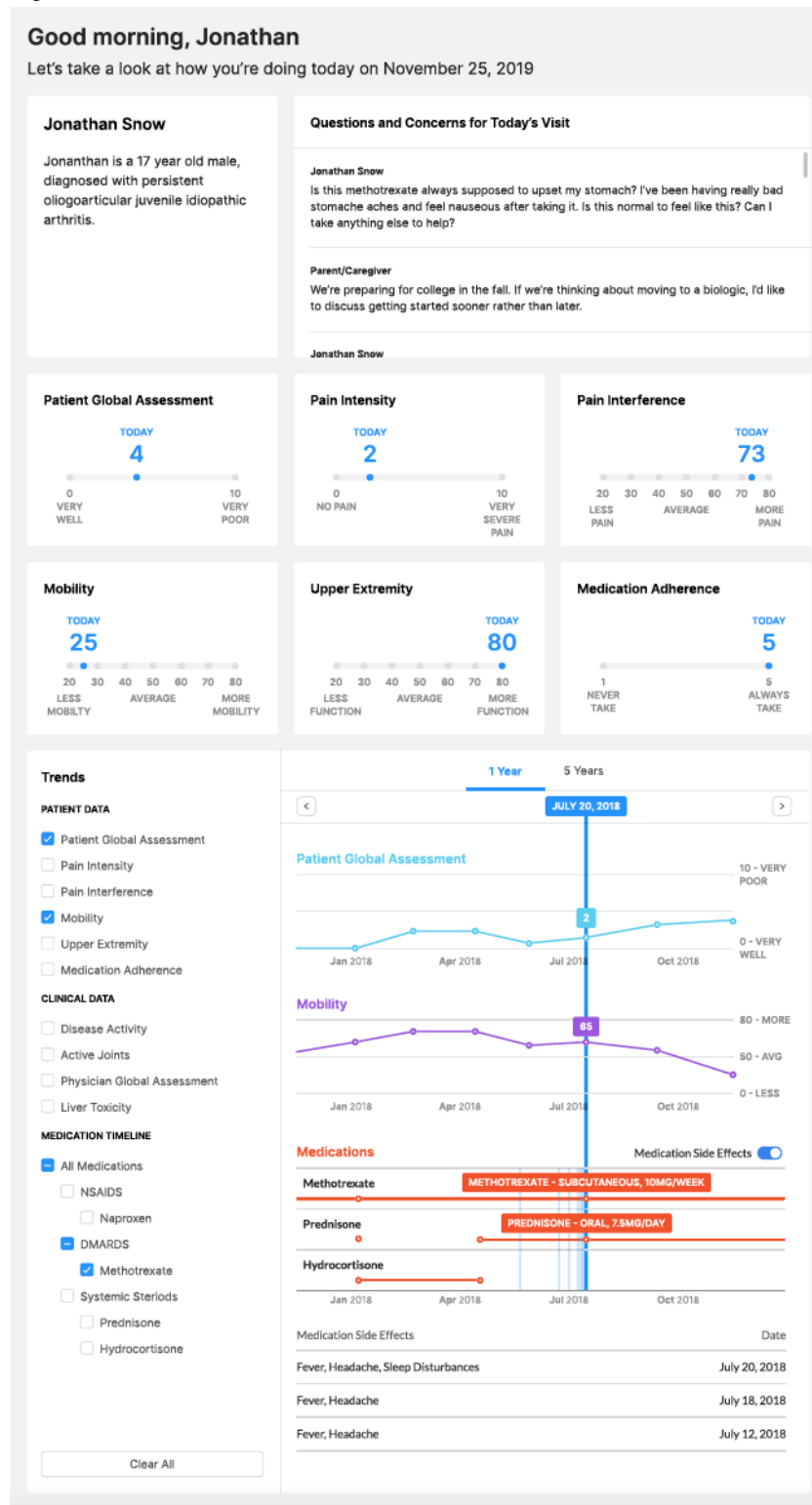
Parents reported that being asked in advance about what they wanted to discuss most prevented them from forgetting anything during the visit. Clinicians appreciated understanding patient and parent concerns and ensured that these questions or concerns were addressed. One clinician shared an experience of how the responses highlighted the specific concerns of both the patient (*scared and did not want to restart medications*) and his parents (*concerned about setbacks from their child's flare*), setting the framework for the visit. Another clinician reported, "We often forget to ask certain questions, and the dashboard *reminds* us to focus on the patient's concerns rather than just looking at a clinical picture."

Feedback on the usability of the paper-based prototype also yielded considerations for the dashboard design and data display, including (1) ensuring that the dashboard is updated on a

real-time basis to include the current visit's clinical assessment data as an important element of visualizing progress over time and engaging in shared decision-making, (2) clearly labeling the graphs for ease of reading and interpretation given varying scales of the data elements, and (3) orientation of the flow vertically to facilitate review and discussion.

The final dashboard design wireframe is illustrated in [Figure 7](#). Three sections of the dashboard emerged: (1) patient and parent and family questions and concerns about the visit, (2) a snapshot of how the patient is currently doing, and (3) a trends section that longitudinally displays PROs, clinical data, and medications and reported side effects. The layout is intended to correspond to the clinical workflow. The top two sections provide the clinician with initial insight into the patient and family needs and how the patient is currently feeling, allowing for more rapid movement into the core of the visit. The trends section is intended for review after data from the clinical assessment have been captured and updated in real time.

Figure 7. Final dashboard design wireframe.



Discussion

Principal Findings

This study used human-centered design principles to involve parents, patients, clinicians, and care team members in the development of a coproduction of care dashboards for clinical use in 4 diverse ambulatory pediatric rheumatology clinics across the United States. This process included evaluating the

context of use and needs of end users, obtaining consensus on necessary data elements, and constructing a display prototype. Notable themes included the right data, in the right place, at the right time; data in once for multiple purposes; patient and family self-management components; and opportunity for education and increased transparency. A final set of 11 dashboard data elements was identified, including PROs, clinical data, and medications. Important design considerations include the incorporation of real-time data, clearly labeled graphs, and

vertical orientation to facilitate reviews and discussions. Prototype paper-testing with 36 patients and families yielded positive feedback about the dashboard's usefulness during clinic discussions, helped to talk about what mattered most, and informed health care decision-making.

Key components of the dashboard display included PROs, clinical data, and medications; all trended longitudinally with clearly labeled graphics and plans for real-time updates. Interestingly, a previsit agenda-setting question to be asked of both patients and families for "Questions or Concerns?" is deemed an important component of patient-centered care. This is consistent with findings in other coproduction projects in which patient and family questions and concerns helped to focus the content of the visits and prioritize what matters most to them [4,6]. In addition, our final dashboard prototype is similar to dashboards developed for adult RA [19] in their longitudinal presentation of PRO and clinical data along with medications, intended to enhance communication about how well medications work to improve patient symptoms, functioning, and disease activity.

Notably, all teams agreed that data collection should be streamlined and used for clinical care, collaborative improvement networks [13], and research, an important step toward the development of an integrated and sustainable learning health system [32]. Our prototype design encompasses the framework of having the right data at the right time to foster enhanced communication and collaboration during a clinical encounter and emphasizes the need for streamlined data collection to support multiple purposes and uses. Many of the dashboard data elements (functional status, pain score, patient global, joint count, and JADAS) were congruent with data collected within the CARRA [12] and PR-COIN [26] registries, as well as the Canadian JIA research network [33]. An opportunity exists to integrate these clinical care data with those required for research and quality improvement purposes. This integration represents a core pillar of building a learning health system [32] and allows for reduced burden on patients and families and more efficient data usage and optimization. Although our design is a paper prototype with an associated wireframe, the long-term goal is to develop an electronic dashboard integrated within the EHR. Notably, because all institutions use the same EHR, we anticipate that the use of standardized data mapping will facilitate interoperability among research networks.

Although prior work has been completed on dashboards in rheumatology [17,19], our study is unique from these published works in several notable ways. First, our study, which focused on the population with JIA, included 4 pediatric rheumatology teams, including patients and parents as team members, from diverse geographic locations who worked collaboratively throughout the co-design process. We also deployed a variety of human-centered design activities, including process mapping, personas, journey mapping, dashboard sketches, and observations. Third, our final set of dashboard data elements was greater than that of the adult population with RA [17,19]. Although both the adult and pediatric rheumatology dashboard data sets include a composite disease activity score, our teams felt that it was important to include component measures

(physician global assessment, tender and swollen joint count, and patient global assessment) to promote greater understanding by patients and families regarding how this score is derived and used. Finally, and most notably, our set includes an agenda-setting question and questions regarding medication adherence and possible side effects to prompt discussion about medication intolerance, which can have a significant impact on quality of life [34].

Strengths and Limitations

The strengths of our study include integrating patients and parents as full members of the pilot site teams, employing a variety of activities to gain insight into the needs of end users, and collecting qualitative and quantitative data to achieve consensus on a dashboard design. We used rapid-cycle iterative testing of a paper dashboard to simulate how a dashboard might work to support the coproduction cycle of (1) coassessing the patient's current health status, (2) codeciding the next steps, (3) co-designing the care plan, and (4) codelivering care [35].

Although this study highlights the importance of involving end users in the design process, we acknowledge several potential limitations in our approach. We leveraged highly engaged clinical sites, care teams, and patients and families and used convenience sampling for development and testing; therefore, the perspectives we gained may not be representative of all pediatric rheumatology practices or populations. We engaged 4 diverse clinical sites, including small and large centers, in various locations across the United States. As we proceed to the next phase of building an electronic version of the dashboard, we will have the opportunity to test the dashboard and assess its usability and utility across a larger target population. Another limitation was the inability to pursue the design of a patient and family self-management tool. Patients and families expressed a strong desire to have a tool for individualized daily symptom tracking and note-taking to capture their experiences of living with a chronic disease. We acknowledge the importance of these functionalities [36]; however, the technological requirements for integrating them as part of a point-of-care dashboard were determined to be beyond the initial scope and capabilities of our study.

Conclusions

We used a human-centered design process to actively engage patients with JIA, families, clinicians, and care teams to successfully create a blueprint for a point-of-care coproduction dashboard to foster meaningful conversations and shared decision-making about care and treatment plans. The necessary dashboard data elements include PROs, clinical data, and medications, and the display should use real-time data, have clearly labeled graphs, and a vertical orientation. Data capture that supports clinical care and research and improvement efforts is ideal. Results from dashboard testing indicated that it was useful during clinical discussions, helped to talk about what mattered most, and informed health care decision-making.

Future study efforts informed by this work and planned by the authors include (1) creating an electronic version of the point-of-care dashboard, (2) preparing for a successful launch through workflow integration and patient and family education

efforts, (3) testing and implementing the dashboard at the 4 pediatric rheumatology pilot sites, and (4) conducting a formative evaluation of its usability and utility in supporting coproduction of care.

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Conflicts of Interest

None declared.

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Abbreviations

CARRA: Childhood Arthritis and Rheumatology Research Alliance
EHR: electronic health record
JADAS: Juvenile Arthritis Disease Activity Score
JIA: juvenile idiopathic arthritis
MoSCoW: must have, should have, could have, will not have
PR-COIN: Pediatric Rheumatology Care and Outcomes Improvement Network
PRO: patient-reported outcome
RA: rheumatoid arthritis
US: United States

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Original Paper

Co-designing Improved Communication of Newborn Bloodspot Screening Results to Parents: Mixed Methods Study

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Abstract

Background: Each year in England, almost 10,000 parents are informed of their child's positive newborn bloodspot screening (NBS) results. This occurs approximately 2 to 8 weeks after birth depending on the condition. Communication of positive NBS results is a subtle and skillful task, demanding thought, preparation, and evidence to minimize potentially harmful negative sequelae. Evidence of variability in the content and the way the result is currently communicated has the potential to lead to increased parental anxiety and distress.

Objective: This study focused on the development of co-designed interventions to improve the experiences of parents receiving positive NBS results for their children and enhance communication between health care professionals and parents.

Methods: An experience-based co-design approach was used to explore experiences and co-design solutions with 17 health professionals employed in 3 National Health Service Trusts in England and 21 parents (13/21, 62% mothers and 8/21, 38% fathers) of 14 children recruited from the same 3 National Health Service Trusts. Experiences with existing services were gathered via semistructured interviews with health professionals. Filmed narrative interviews with parents were developed into a composite film. The co-design process identified priorities for improving communication of positive NBS results through separate parent and health professional feedback events followed by joint feedback events. In total, 4 interventions were then co-designed between the participants through a web-based platform.

Results: Parents and health professionals provided positive feedback regarding the process of gathering experiences and identifying priorities. Themes identified from the parent interviews included impact of initial communication, parental reactions, attending the first clinic appointment, impact of health professionals' communication strategies and skills, impact of diagnosis on family and friends, improvements to the communication of positive NBS results, and parents' views on NBS. Themes identified from the health professional interviews included communication between health professionals, process of communicating with the family, parent- and family-centered care, and availability of resources and challenges to effective communication. In response

to these themes, 4 interventions were co-designed: changes to the NBS card; standardized laboratory proformas; standardized communication checklists; and an email or letter for providing reliable, up-to-date, condition-specific information for parents following the communication of positive NBS results.

Conclusions: Parents and health professionals were able to successfully work together to identify priorities and develop co-designed interventions to improve communication of positive NBS results to parents. The resulting co-designed interventions address communication at different stages of the communication pathway to improve the experiences of parents receiving positive NBS results for their children.

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KEYWORDS

experience-based co-design; neonatal screening; health communication; participatory research

Introduction

Background

Newborn bloodspot screening (NBS) in England involves collecting a small sample of blood on a special card from a baby's heel on day 5 of their life. This is then sent to an NBS laboratory to be analyzed. Positive NBS results are reported to relevant clinical teams, often using locally developed proformas [1] who then communicate the result to the family. Each year in England, almost 10,000 parents are informed of their children's positive NBS results approximately 2 to 8 weeks after birth depending on the condition [2,3]. The purpose of NBS is to identify presymptomatic babies affected by one of 9 conditions currently screened for to enable treatment to be initiated early to improve outcomes for the child. The conditions are sickle cell disease (SCD), cystic fibrosis (CF), congenital hypothyroidism, phenylketonuria, medium-chain acyl-CoA dehydrogenase deficiency, maple syrup urine disease (MSUD), isovaleric acidemia, glutaric aciduria type 1, and homocystinuria (pyridoxine-unresponsive)—the latter 6 collectively referred to as inherited metabolic diseases. The clinical spectrum in screen-positive cases varies enormously and, consequently, the message to parents needs to be carefully crafted to prepare for a range of outcomes.

Communication of positive NBS results is a subtle and skillful task that demands thought, preparation, and evidence to minimize potentially harmful negative sequelae [4-8]. For instance, the perceived lack of knowledge of the person communicating the NBS result rather than the actual result has been linked to parental distress [4]. Poor or inappropriate communication strategies for positive NBS results can also influence parental outcomes in the short term [4-7,9,10] but may also have a longer-term impact on children and families [8]. Evidence suggests that the distress caused can manifest in several ways, including arguments between couples, apportioning of blame [4,6,11], alteration of life plans and inability to conduct tasks of daily living such as going to work or socializing [4], long-term alterations in parent-child relationships [8], and mistrust and lack of confidence affecting ongoing relationships with health care staff [6]. There is also evidence of increased parental concern resulting in parents reducing their child's interaction with others, particularly in the case of CF [4]. Parents also experience poor intra- and

interpersonal relationships within their family systems and more widely [12].

This supports the importance of ensuring that the initial communication of positive NBS results is handled sensitively and considers individual parent characteristics to minimize parental distress and the consequences of this distress, as well as the knowledge and experience of the person imparting the result. The choice of approach is, to some extent, influenced by the seriousness of the condition identified and the need for an immediate or less immediate response. In one study, parents who had received the screening results from a CF specialist were more satisfied than those who had received the screening results from the maternity ward [13]. In another study, information received by telephone was less satisfactory to parents of children diagnosed with CF (odds ratio 2.23; $P=.04$) or parents of younger infants (odds ratio 0.93 per day older; $P=.001$) [10]. Results delivered over the phone by staff not known to the families or without condition-specific knowledge were viewed less favorably and contributed to parental dissatisfaction, anxiety, and distress [9].

Recognizing the need to work with parents and health professionals to improve this communication, the *Rethinking Strategies for Positive Newborn Bloodspot Screening Result Delivery: a process evaluation of co-designed interventions* project sought to develop, implement, and evaluate new interventions to improve the delivery of initial positive NBS results to parents. This mixed methods study comprised 3 main phases. Phase 1 involved a national survey using telephone interviews to explore current approaches to the communication of positive NBS results [14] and inform the selection of 2 study sites for the remaining phases. The second phase used the principles of experience-based co-design (EBCD) to explore health professionals' and parents' experiences of delivering and receiving positive NBS results, respectively. Findings from interviews with health professionals have been published elsewhere [1]; sections of this paper related specifically to these findings have been reproduced from *BMJ Open* under license CC-BY-4.0. In addition, EBCD was used to develop interventions for communicating positive NBS results to parents. In phase 3, the interventions were evaluated in 2 selected case study sites (2 NBS laboratories that served 3 National Health Service [NHS] Trusts in England) [15].

Aim

The aim of the research reported in this paper was to describe the use of a modified version of EBCD during phase 2 to develop co-designed interventions to improve the experiences of parents receiving positive NBS results for their children and enhance the communication between health care professionals and parents.

Methods

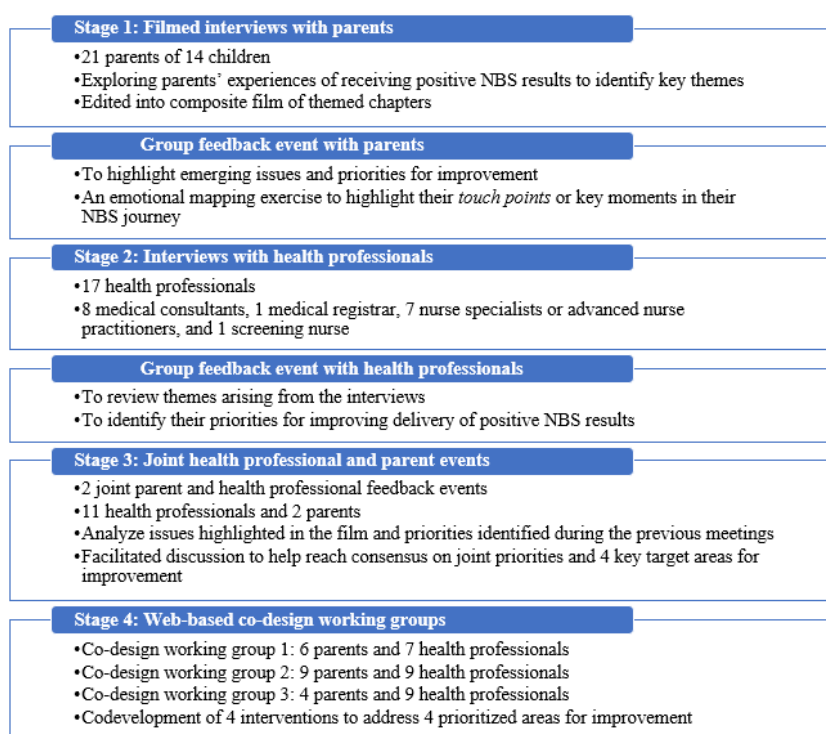
Overview

This formative study was underpinned by family systems theory (FST) [16] because of the potential vulnerability of family relationships if the initial positive NBS result information is not shared as effectively and empathetically as possible [17]. In FST, all components of the family are regarded as interdependent—what happens to one member will affect all other members of the family directly and indirectly. FST postulates that family functioning has the potential to be affected by an event such as the communication of the initial positive NBS result and, subsequently, facilitating the coping mechanisms used and the adaptation of families to the NBS result is paramount.

The co-design process was informed by the EBCD toolkit [18]. EBCD was selected because of its focus on service users and health professionals working in partnership to develop and improve health services. This was felt to be particularly appropriate as family-centered care, which includes working in partnership with the family, is the principal philosophy of pediatric care in many countries worldwide [19]. EBCD is an approach to improving health care services that draws on participatory design and user experience to bring about quality

improvements in health care organizations [20]. This involves focusing on and designing patient or carer experiences rather than just systems and processes [21-23]. The co-design process enables staff, patients, and carers to reflect on their shared experiences of a service and then work together to identify improvement priorities, devise and implement changes, and then jointly reflect on their achievements. EBCD was piloted in an English head and neck cancer service in 2005 [21]. After a subsequent project in an integrated cancer unit, a web-based toolkit [18] was developed as a free guide to implement the approach. A recent systematic review identified 20 studies that had used EBCD mainly in mental health and cancer services in the United Kingdom. This review highlighted variations in the use of EBCD, with many of the studies eliminating or modifying some of the EBCD stages. It has been recognized that the disadvantages of EBCD include it being time-consuming and expensive. Until recently, EBCD had mainly been used with adult service users or their carers or family members. The use of EBCD with parents with or without the participation of their children is still quite novel, having only been explored more recently and with adaptations to the process [24-26]. Therefore, this study also builds on knowledge of using this method with parents.

The EBCD process was modified to gather parents' and health professionals' experiences and agree on areas for improvement in the communication of positive NBS results to families. It followed four stages (Figure 1): (1) engaging health professionals and gathering experiences (the findings from health professional interviews have been published elsewhere [1]), (2) engaging parents and gathering their experiences, (3) bringing parents and health professionals together to share experiences and identify priorities for improvement, and (4) web-based co-design activities.

Figure 1. Adapted experience-based co-design approach. NBS: newborn bloodspot screening.

Patient and Public Involvement

Patient and Public Involvement (PPI) was instrumental in the design and conduction of this study. A total of 8 parents of babies who had received a positive NBS result for one of the 9 screened conditions formed a PPI group that met every 6 months for the duration of the study. Their suggestions were incorporated into the study design, data collection tools, and data analysis and dissemination. The PPI group was presented with data from the annual reports of the NBS programs and made suggestions as to which sites should be used during the co-design process. In addition, views of representatives from charities for the screened conditions, including Metabolic Support UK, the British Thyroid Foundation, the CF Trust, and the Sickle Cell Society, were also incorporated.

Study Sites and Sampling

Overview

The study sites consisted of 3 NHS provider organizations (Trusts) in England served by 2 NBS laboratories (study sites) that process comparable numbers of positive NBS reports annually for each of the 9 conditions currently included in the NBS program. These consisted of 2 Trusts in Greater London served by 1 NBS laboratory and 1 NBS laboratory in the West Midlands that processed 128 and 129 positive NBS results, respectively, in 2017 to 2018.

Informed by previous successful EBCD projects [20,22,27], we recruited a purposeful sample of parents across the 2 study sites. This ensured the participation of parents who (1) had received a positive NBS result for their child (2) in the previous 3 to 36 months, as well as ensuring (3) the representation of all screened conditions. Parents were identified as potential participants by

health professionals communicating positive NBS results. During a routine hospital appointment, health professionals asked parents if they would be willing to talk to a member of the research team about the study. If the parents agreed, a member of the research team met with them, explained the study, and provided a participant information sheet. Parents were asked if they would be willing to share their contact details so that a member of the research team could contact them the following week to answer any questions they might have about the study. During the follow-up contact, if parents were agreeable, an appointment was made to undertake the filmed interview at a convenient time and location of the parents choosing (all parents chose to be interviewed at home).

A 2-stage sampling approach was used to recruit health professionals involved in communicating positive NBS results in the preceding 6 months at the 2 study sites. Participants were first sampled purposively based on their experience with reporting or communicating positive NBS results, followed by a second stage of snowball sampling. Members of relevant clinical teams (medical consultants, general pediatricians, nurse specialists, and specialist screening nurses) were initially identified through individual Trust websites, contacted via email, and invited to participate. If no response was received, a follow-up email was sent after 1 week. Health professionals who responded were asked if there were any other members of their clinical teams that the research team should contact to ensure that the views were representative.

The sample sizes for both parents and health professionals were influenced by previous EBCD projects and the EBCD toolkit [18].

Stage 1: Engaging Parents and Gathering Experiences

Participants

Filmed interviews were conducted with 21 parents: 13 (62%) mothers and 8 (38%) fathers of 14 children recruited from 3 NHS Trusts in England served by 2 NBS laboratories. Of the 21 parents, 18 (86%) identified as White British, 1 (5%) identified as White European, 1 (5%) identified as Asian British, and 1 (5%) identified as Black British. Their ages ranged from 25 to 44 (median 37) years. Of the 14 children, 4 (29%) had CF; 3 (21%) had medium-chain acyl-CoA dehydrogenase deficiency; 2 (14%) had phenylketonuria; 1 (7%) had MSUD; 1 (7%) had congenital hypothyroidism; 1 (7%) had SCD; 1 (7%) had been designated CF screen-positive, inconclusive diagnosis; and 1 (7%) had received a false positive result for CF. Of the 14 children, 7 (50%) had older siblings, only one of whom had also been diagnosed with a condition (CF) via NBS. A total of 14% (2/14) of the children were twins (both had CF), and 36% (5/14) did not have any siblings. At the time of the interview, the ages of the children ranged from 10 to 107 (median 43) weeks.

Data Collection

We conducted filmed narrative interviews with parents across the 2 study sites between September 2018 and March 2019 exploring parents' experiences of receiving positive NBS results to identify key themes (touch points). Interview questions were guided by the principles of FST [16,17] and focused on the impact of receiving a positive NBS result on the family and on their relationships with each other, with their children, and also with their wider support network, including their friends. The interviews lasted between 14.5 and 47.4 (median 26.4) minutes. Parents were asked to talk about their experience of receiving their child's positive NBS result both in terms of the process and any emotions or feelings this had caused and why.

Textbox 1. Prompts for the parent feedback event.

Key questions

- Do you feel the film represents your views and experiences?
- What parts of your journey were you happy with? Why?
- What parts of your journey do you think could be improved? How?
- What questions would you like to ask health professionals?

Stage 2: Engaging Health Professionals and Gathering Experiences

Participants

Health professionals were recruited from the same 3 NHS Trusts in England served by 2 NBS laboratories. In total, 20 health professionals involved in communicating positive NBS results in the preceding 6 months were emailed and invited to participate, of whom 2 (10%) did not respond to the invitation and 1 (5%) did not communicate the initial positive screening result and was therefore ineligible. In line with the EBCD approach [18], 16 face-to-face interviews were conducted with 17 health professionals (2/17, 12% requested to be interviewed together), of whom 8 (47%) were from one of the NBS

Data Analysis

FST [16,17] informed the development of themes identified from parent interviews. This included consideration of parent reactions to receiving the positive NBS result and of how this had affected them as parents, individuals, and partners, as well as the impact of the diagnosis on family and friends, reflecting the tenets of holism and interdependence that are fundamental to FST. Themes identified from parent interviews were developed into a composite film in April 2019. The film was used to capture parents' experiences of receiving their children's positive NBS results and provide rich information to guide the development of the co-designed interventions.

Following the interviews, parents at each study site were invited to a parent feedback event (1 in the West Midlands and 1 in London) to enable them to watch the composite film and discuss key priorities to improve the communication of positive NBS results to families. These events were guided by the web-based EBCD toolkit [18] and accompanying web-based resources, including the invitation, agenda, and feedback templates. Parents were invited to view the composite film of the interviews to ensure that it was a fair and valid representation of their shared experiences. This was used to inform a facilitated group discussion that lasted approximately 3 hours to highlight emerging issues and priorities for improvement. In addition, an emotional mapping exercise was conducted to highlight their *touch points*, or emotionally charged or key moments in their NBS journey. During this discussion, parents were asked to work together to consider 4 key questions (Textbox 1).

Touch points were gathered from the composite film and the emotional mapping exercise to highlight priorities to share with health professionals.

laboratories and the remaining 9 (53%) were split across the 2 Greater London Trusts served by the other NBS laboratory. Participants with experience with all 9 screened conditions were included. The sample consisted of 47% (8/17) medical consultants, 6% (1/17) medical registrars, 41% (7/17) nurse specialists or advanced nurse practitioners, and 6% (1/17) screening nurses. The length of experience with NBS ranged from 2 to 38 (median 8) years. The interviews lasted 37 minutes on average (SD 10.51, range 19-58 minutes) [1].

Data Collection

Semistructured telephone interviews comprising closed- and open-ended questions were conducted between September 2018 and February 2019 to identify the approaches used to communicate positive NBS results from NBS laboratories to

health professionals. Data collected included the mode of communication strategy (face-to-face, letter, telephone, or email), the resources involved in each communication strategy, who provided the information and their role, and the location (colocated or alternative site) of relevant services for each condition.

After the interviews, health professionals at each site were invited to attend a health professionals' event to review themes arising from the interviews and identify their priorities for improving the delivery of positive NBS results (1 in the West Midlands and 2 in London). These events were guided by the

web-based EBCD toolkit [18] and the accompanying web-based resources, including the invitation and agenda template. The findings of the health professional interviews were presented via a Microsoft PowerPoint presentation using direct quotes to illustrate the points made. The participants were encouraged to reflect on what they considered to be working well, what they thought required improvement and, from this, key priorities to improve the communication of positive NBS results to families. Health professionals were asked to record their thoughts on a flip chart paper so it could be shared with the whole group (Figure 2).

Figure 2. Illustrative flip charts from health professional workshops.



Data Analysis

The interviews were analyzed thematically; an inductive method of data analysis was used, and themes were generated using a latent approach. This provided a deeper understanding of the approaches used to communicate positive NBS results to families [28]. In total, 2 members of the research team (JC and HC) coded 1 interview transcript separately. These codes were then compared to inform and align code development [29], and a codebook was developed [30]. A further 4 transcripts were then coded separately by the same 2 members of the research team using the codebook. These separately coded transcripts were then compared; intercoder reliability was 84%. Following this, the same 2 members of the research team coded the remainder of the transcripts using the codebook. Once this initial coding had been completed, data for each code were compared to ensure consistency in coding and to enable the codes to be collapsed into themes. All quotes for each theme were collated to inform theme development. This was an ongoing, iterative process; new codes were developed, and the definition of codes was refined as the analysis progressed [1].

Stage 3: Bringing Health Professionals and Parents Together

Participants

Health professionals and parents who had taken part in the previous events were invited to take part in one of 2 joint parent-health professional feedback events: 1 in the West Midlands and 1 in London. A total of 6 health professionals and 1 parent joined the event in the West Midlands, and 5 health professionals and 1 parent joined the London event.

Data Collection

Mixed health professional and parent events [31] were held at each of the study sites. These events were face-to-face and took approximately 2 to 3 hours. These events were guided by the web-based EBCD toolkit [18] and the accompanying web-based resources, including the invitation and agenda template. During these events, a parent representative (discussed and agreed upon before the meeting) was invited to introduce and share the composite film with health professionals. An unstructured discussion followed to analyze issues highlighted in the film and priorities identified during the separate health professional and parent meetings. This was followed by a facilitated discussion to help reach a consensus on joint priorities. In total, 4 key target areas for improving the delivery of positive NBS results [20,27,32] were agreed upon to be the focus of the co-design activities over the following 8 weeks (July 2019 and August 2019).

Data Analysis

During the joint health professional and parent feedback event, the participants were asked to write on Post-it notes placed on flip chart paper what they currently considered to be working well, what areas they thought needed improvement, and priorities. These were shared with the group and, following a facilitated group discussion, shared priorities were agreed upon, and key target areas were identified for improvement of communication of positive NBS results to parents.

Stage 4: Co-design Working Groups

Participants

A total of 3 co-design working groups were run, each attended by 12 to 18 participants (Figure 1). The participants were permitted to be part of more than one co-design working group if they wished.

Data Collection

The co-design working groups took place in July 2019 and August 2019. EBCD is typically undertaken through face-to-face events [18]. It was modified in this case as health professionals and parents requested that the co-design working groups be held on the web. The rationale for this was to offer more flexibility to share resources but also to facilitate communication and negotiation between health professionals and parents regarding the proposed co-designed interventions.

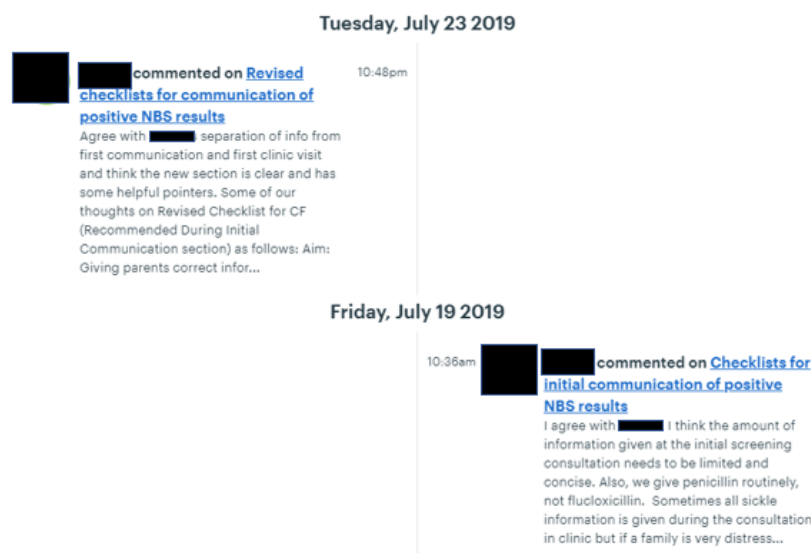
The web-based platform Basecamp [33] was used to host the web-based co-design working groups. Each co-design working group was set up as a different group; those who had indicated that they would be interested in a particular co-design working group were invited via email to participate.

Ground rules were jointly agreed upon at the outset and posted on the web. The *Message Board* was used to invite participants (a mixture of health professionals and parents in each co-design working group) and remind them of the purpose of the groups. The composite film as well as Microsoft PowerPoint presentations and priorities from the separate and joint parent and health professional events were made available. Example interventions based on discussions held during the separate and joint parent and health professional feedback events were also shared, and members of the co-design working groups were

asked to provide feedback and comments. The *Campfire* function was used for discussion related to iterations of all documents. Each time new documents were uploaded, a message was sent to the members of the relevant co-design working group via the *To-dos* function.

The participants were asked, over a period of 8 weeks from July 2019 to August 2019, to post comments on documents and files that were uploaded. Members of each group were sent a message approximately weekly or when new or revised documentation was uploaded to the web-based portal asking them to review the information and provide feedback. They also used the web-based discussion board to communicate with each other and develop the co-designed interventions. An example of communication between parents and health professionals through this platform is shown in Figure 3. Versions of relevant documents were updated in light of health professionals’ and parents’ comments until a consensus was reached regarding the suitability of the proposed interventions. Both parents and health professionals engaged effectively with the web-based co-design working groups. Comments and feedback were left at all times of the day and night, indicating that using the web-based forum enabled participants to contribute to the co-design working groups at times that were convenient for them. Conducting the co-design working groups on the web also appeared to mitigate any potential imbalance in terms of perceived power hierarchies between parents and health professionals [34], with both contributing and replying to each other’s comments. Furthermore, being able to monitor which participants had contributed comments or feedback meant that it was easier to direct questions to participants who had been less forthcoming in discussions and encourage their involvement in a nonconfrontational manner.

Figure 3. Redacted example of communication during the co-design working groups.



Ethics Approval

All potential participants were given the choice to take part or not and were reminded of their right to withdraw from the study at any time. Written informed consent was obtained from all participants. This study is part of a larger program of work [35]

and was approved by the London – Stanmore Research Ethics Committee (17/LO/2102).

Results

Experiences and Views

A total of 5 themes were identified from the interviews with health professionals: communication between health professionals, process of communicating with the family, parent- and family-centered care, and availability of resources and challenges to effective communication. Data from the interviews with health professionals have been published in full elsewhere [1].

Themes identified from the interviews with parents included impact of initial communication, parental reactions, attending

the first clinic appointment, impact of health professionals' communication strategies and skills, impact of diagnosis on family and friends, improvements to the communication of positive NBS results, and parents' views on NBS. The findings were presented as a composite film (available via the study blog [36]) to capture and illustrate parents' experiences of receiving their children's positive NBS results and provide rich information to guide the co-design activities. The film is presented in 7 sections that reflect the stages of parental experiences and their journeys through screening. The common experiences or *touch points* for parents that were reflected in each section of the film are summarized in [Textbox 2](#).

Textbox 2. Touch points from the composite film.

Section 1: initial communication

- Various methods of communication were used including face-to-face, telephone, and SMS text message.
- The characteristics of the person communicating the newborn bloodspot screening (NBS) result were important.
- The person communicating the NBS result was not always knowledgeable about the condition and could be viewed as unreliable.
- Mothers frequently communicated the result to their partners.
- The NBS result was perceived to be delivered as "bad news," which contributed to their initial feelings of fear and pain (see below).

Section 2: parents' reactions

- Common feelings: shock, fear, confusion, pain, and disbelief
- The positive NBS result was traumatic, upsetting, and devastating.

Section 3: attending the first clinic appointment

- The wait between the initial communication and the first clinic appointment was difficult (this was normally <24 hours).
- Practical arrangements had to be made at short notice (eg, travel, which could be expensive, and childcare for other children).
- The initial clinic appointment was exhausting.

Section 4: health professionals' communication

- Condition-specific specialists were found to be positive, supportive, knowledgeable, empathetic, reassuring, and credible.

Section 5: impact of diagnosis on family and friends

- Some parents reported that the positive NBS result had brought them closer together.
- Some felt it had created a strain on their relationship.
- Some felt it had affected their relationship with their baby in terms of bonding and attachment.
- Parents felt responsible for telling family and friends.

Section 6: improvements to the communication of positive NBS results

- Those involved should be knowledgeable about the conditions and the process when communicating positive NBS results.
- Partners should be informed at the same time as mothers.
- An SMS text message alert (or similar) could help prepare parents to receive the positive NBS result.
- The NBS result should be communicated to parents by a condition-specific specialist.
- Information should be provided immediately after the child's positive NBS result is relayed.

Section 7: parents' views on NBS

- The NBS program was viewed very favorably.
- New parents should be encouraged to participate in the NBS program.
- Midwives should be familiar with the conditions included in NBS.

Priorities for Improving Communication

During a facilitated discussion after watching the film of parental experiences, feedback from parents and health professionals

was narrowed down to a short list of priorities for them to explore together to improve communication. These are summarized in [Table 1](#).

Table 1. Summary of participants’ priorities to improve communication.

Category	Parents’ priorities	Health professionals’ priorities
Changes to NBS ^a card	<ul style="list-style-type: none"> • How the parent would like to be contacted • Significant other’s contact details on the card (as well as the mother’s) • Whether a translator is needed • Email address of the parents 	<ul style="list-style-type: none"> • Inclusion of a question on the NBS card asking the parents how they would like to be contacted: Skype, telephone, or email • Addition of a parent’s email address to the NBS card
Initial communication	<ul style="list-style-type: none"> • Being told by the same person they will see at the first clinic appointment • If parents are given their child’s result over the telephone, care should be coordinated so that they can speak to a health visitor (registered nurses or midwives who have undertaken additional training and work mainly with children from birth to 5 years and their families) or midwife after for support (they do not need to have knowledge of the condition) • Parents to be told who they can or should bring to the first clinic appointment 	<ul style="list-style-type: none"> • Templates for communication to clinical teams and initial communication to families that should be condition specific • Information for families about who should attend the initial clinic appointment
Follow-up communication	<ul style="list-style-type: none"> • Parents to be emailed details of the first clinic appointment • Information for family and friends • Being signposted at this stage with trustworthy and reliable resources or websites 	<ul style="list-style-type: none"> • Following delivery of the positive NBS result by phone, email parents with appointment letter, directions, and condition-specific leaflet; this can be done by administrators or the CNS^b • Information resources for families and extended families
Service provision	<ul style="list-style-type: none"> • Financial support for families to attend the initial clinic appointment 	<ul style="list-style-type: none"> • A centralized system for CHT^c • Formulation of diagnostic services especially out of hours (so laboratories can conduct confirmatory testing over the weekend) • Financial support for families to attend the initial clinic appointment

^aNBS: newborn bloodspot screening.

^bCNS: clinical nurse specialist.

^cCHT: congenital hypothyroidism.

Co-design Working Groups and Interventions

During the joint parent and health professional groups, the participants narrowed down the initial priorities ([Table 1](#)). Through discussions and shared expertise on the potential causes of communication issues, they decided on the focus of each of the co-design working groups. This is summarized in [Table 2](#).

The participants agreed that changes to the NBS card (completed during the heel prick test by the midwife) were required to address the challenge of having all the information necessary to contact the family (1) in a timely (condition-specific) manner and (2) according to parental preferences.

There was also a focus on standardized laboratory proformas for use in the NBS laboratories. This focus emerged from a need for consistent and thorough information to be relayed to clinical teams to facilitate making contact with the child’s family following a positive NBS result.

Parents recognized inconsistent communication approaches. It was agreed that standardized communication checklists for

health care professionals would guide conversations throughout the screening journey and support health professionals with less condition-specific knowledge or experience.

A template email or letter to the parents was proposed as the fourth intervention. This would be sent by the clinical team after the initial communication with the parents. The purpose would be to provide reliable up-to-date, condition-specific information for parents following the communication of the positive NBS result.

Through the co-design process, ideas and documentation were reviewed and iterated through the Basecamp platform until a consensus was reached regarding the suitability of the proposed interventions. Overall, there were 6 iterations of the NBS card, 5 iterations of the laboratory proformas, 8 iterations of the communication checklists, and 6 iterations of the email or letter for providing information to parents following the communication of the positive NBS result. Examples of the final versions are outlined in the following sections.

Table 2. Co-design working groups (CDWGs).

Group	Proposed intervention	Need
CDWG 1	<ul style="list-style-type: none"> Changes to the NBS^a card completed during the heel prick test by the midwife Standardized laboratory proformas for use in the NBS laboratories 	<ul style="list-style-type: none"> To ensure that health professionals have all the required information to make rapid contact and that parents are contacted in their preferred way To ensure that the required information is consistently transferred from the laboratories to clinical teams
CDWG 2	<ul style="list-style-type: none"> Standardized communication checklists for health care professionals 	<ul style="list-style-type: none"> To ensure that the required information is relayed consistently to families during the initial communication
CDWG 3	<ul style="list-style-type: none"> A template email or letter to parents 	<ul style="list-style-type: none"> To provide reliable, up-to-date, condition-specific information for parents following the communication of the positive NBS result

^aNBS: newborn bloodspot screening.

The NBS Card

The final version of the proposed NBS card included the addition of parents' preferred method of contact. This aimed to prompt conversation between midwives and parents at the time the NBS sample was taken regarding the possibility of them being contacted in the future if the results were positive as well as to ensure that parents were involved in the decision about how they might be contacted. Alternative contact details of a

significant other were also added to act as a second line of contact should a clinician be unable to reach the mother following the NBS result. The parents' email addresses were added to aid future communication and contact. Finally, the option to add information related to any hearing or sight impairments or language needs that might hinder future communication with parents was added to the NBS card. The changes and additions are shown in [Figure 4](#).

Figure 4. New newborn bloodspot screening card.

Standard Laboratory Proformas

The standard laboratory proformas built on those developed by the Department of Clinical Chemistry and Newborn Screening at the Sheffield Children's NHS Foundation Trust. The proformas were condition specific and included a front page that was mainly intended for completion by the NBS laboratory and a section for completion by the clinicians to be fed back to

the NBS laboratory. On the reverse side, there was a reminder of the current referral guidelines, more information about the child's NBS result, and a checklist focused on steps in the referral process. Additions as a result of the co-design process included information related to recommended actions following a positive NBS result for each condition and a comment section to allow clinicians to record suggested condition-specific relevant information ([Figure 5](#)).

Figure 5. Example co-designed laboratory proforma for cystic fibrosis.

Name of Lab:
Contact details/Phone number:

NOTIFICATION OF CONDITION SUSPECTED FROM NEWBORN SCREENING – CYSTIC FIBROSIS		
CONFIDENTIAL – PATIENT INFORMATION		
Referred by (name and designation):	Date:	Case/ laboratory ID:
Tel:	E-mail:	
Referred to (name and designation):	Location:	
Tel:	E-mail:	
Resources e.g. parent leaflet, communication, diagnostic and treatment guidelines available at: https://www.gov.uk/government/collections/newborn-blood-spot-screening-programme-supporting-publication#cystic-fibrosis-cf		
PATIENT DETAILS		
NHS Number:	Date of birth:	Gestation:
Name:	Gender:	Location: Home/Hospital
Gestation:	Birth weight (g)	
Address:	Telephone number(s):	
Post Code:		
Mother's Name:	Mother's date of birth:	
Mother's NHS number:		
TEST RESULTS:		
Initial sample:	Date: DD/MM/YY	IRT (ng/mL whole blood): (cut off =)
Mutation analysis: (including legacy names)		
Second sample:	Date: DD/MM/YY	IRT (ng/mL whole blood): (cut off =)
Disorders from the other eight newborn screening tests (PKU, MCADD, IVA, GA1, MSUD, HCU, CHT and SCD) were: **Not Suspected / In Progress (Please pass information to parents).		
COMMENTS:		
GP (address and phone number):	Consultant:	
Disorders from the other eight newborn screening tests (CHT, PKU, MCADD, IVA, GA1, MSUD, HCU, and sickle cell disease) were: **Not Suspected / In Progress (Please pass information on to parents).		
COMMENTS:		
REQUIRED ACTION please complete and return by secure e-mail to INSERTEMAIL@nhs.net		
Acknowledge receipt of referral (name and designation):		
Date of planned clinic appointment for PP (ideally day after referral):		

Name of Lab:
Contact details/Phone number:

Parents informed by (name and designation):		Date:
Recommended action as per CF Screening Programme Guidelines for Clinical Referral (available from www.newbornbloodspot.screening.nhs.uk/cf/):		
<ul style="list-style-type: none"> unless the primary care team has significant concerns, only inform the family of a positive screening result on a Monday, Tuesday or Wednesday. This avoids the diagnostic assessment being undertaken on a Friday or the family waiting over a weekend for the assessment offer parents an appointment for the diagnostic assessment the following morning the baby must be seen within five working days of the Regional CF Centre being informed of a positive result a sweat test is essential and should occur at the diagnostic assessment visit if possible. This should include measurement of sweat chloride and be undertaken according to ACB standards (www.rcpch.ac.uk/improving-child-health/clinical-guidelines-and-standards/endorsed-and-supported/inherited-metabolic/). If unsuccessful at the first visit, repeat the sweat test at a later stage if a sweat test is not undertaken at the diagnostic assessment, or insufficient sweat is collected, organise repeat CFTR gene analysis at the diagnostic assessment. This can be undertaken on a blood sample or buccal (mouth) swab the diagnostic assessment should include a clinical assessment of the infant 		
Case / laboratory ID:		
TEST RESULTS		
Initial sample:	Date:	Age:
	Mean: IRT (ng/mL): (cut off =)	
	DNA 4 mut:	Further mut analysis:
	Second sample required: Yes / No	Reason: N+/ above action limit 2
Repeat sample:	Date:	Age:
	IRT (ng/mL):	Mean:
PRESUMPTIVE POSITIVE (PP) (e-mail notification to clinical team)		
Referred to (name and designation):		
Email and telephone number:		
PAPERWORK		
DNA report received:	<input type="checkbox"/>	Consultant informed (name and contact number):
CF team informed	<input type="checkbox"/>	E-mail notification of PP form: <input type="checkbox"/>
Copy of card attached to referral:	<input type="checkbox"/>	Result uploaded to CHIS: <input type="checkbox"/>
COMMENTS e.g. siblings with same condition, inpatient location, has baby had penicillin, has baby had any immunisations:		

Communication Checklist

The communication checklists were initially intended to focus on the initial communication of the positive NBS result. However, during the co-design working groups, the participants indicated that they would like the checklists for each stage of the families' NBS journey to include the initial communication (Figure 6), the initial clinic visit, and subsequent clinic visits. It was thought that this would enable all information about the child and family's NBS journey to be recorded in one place. This would also act as an aide-memoire for subsequent clinicians

when seeing the child and family and mitigate the need for parents to recount their story to different clinicians. The initial communication checklists were built on those developed by the CF teams at Sheffield Children's Hospital and King's College Hospital and the Newborn Screening Team at Birmingham Children's Hospital to include more detailed condition-specific information as well as optional information that could be included if deemed appropriate. The checklists for subsequent clinic visits were developed with clinical teams and parents during the co-design process.

Figure 6. Example communication checklist for a child with suspected cystic fibrosis by newborn bloodspot screening.

Name of child:			
Name of person communicating result:	Sign _____	Print _____	
Profession of person communicating result:	Consultant <input type="checkbox"/> Nurse <input type="checkbox"/> GP <input type="checkbox"/> HV <input type="checkbox"/> Other _____		
Method of communication:	Home visit <input type="checkbox"/> Telephone <input type="checkbox"/> Other _____		
RECOMMENDED DURING INITIAL COMMUNICATION OF POSITIVE NBS RESULT			
		Date	Initial
Introduction	Who you are and where you're from (if two parents present, speak to both)	DD/MM/YY	
Check who you are speaking to	Confirm you are speaking to the parents /legal guardians of the baby	DD/MM/YY	
Check correct baby	Name _____ DoB: _____ DD/MM/YY	DD/MM/YY	
Reason for visit / call	Remind parents baby had 'heel prick' when 5 days old	DD/MM/YY	
	One of the results has come back suggesting one of the conditions is suspected	DD/MM/YY	
	Name of the condition _____	DD/MM/YY	
	Not diagnostic, a screening test	DD/MM/YY	
	Need more tests to confirm the result	DD/MM/YY	
	Give date and time of first clinic appointment Date: DD/MM/YY Time HH: MM	DD/MM/YY	
Initial information	Explain that there is a DNA result and this is an inherited condition	DD/MM/YY	
	Ask if they know of any family history	DD/MM/YY	
	Not caused by anything the parents did before or during pregnancy	DD/MM/YY	
	Reassure parents that their baby is well and it is safe to wait until they are seen by clinical team	DD/MM/YY	
	Advise parents to write down any questions they think of so they can ask these at their clinic appointment	DD/MM/YY	
	If face-to-face, give information sources and appointment details	DD/MM/YY	
	Suggest parents to come together or bring someone with them to the appointment	DD/MM/YY	
	Give contact name and number of member of clinical team	DD/MM/YY	
	Give PHE 'suspected' leaflet	DD/MM/YY	
	Discuss suitable websites if appropriate	DD/MM/YY	
Afterwards	If not face-to-face, send email with appointment details, contact information and information sources	DD/MM/YY	
Optional information (If confident and qualified to discuss and if parents are interested in hearing more)	Abnormal protein from abnormal gene	DD/MM/YY	
	Results in altered movement of salt	DD/MM/YY	
	Leads to production of abnormal secretions	DD/MM/YY	
	May also affect digestion – so a stool sample will be collected and sent away	DD/MM/YY	
	Lifelong condition	DD/MM/YY	
	In the UK, around one in every 2500 newborn babies have CF	DD/MM/YY	
Comments		DD/MM/YY	

Email or Letter Template

The email or letter template was intended to be sent to parents immediately after the initial communication of the positive NBS result. These built on those developed by the pediatric metabolic clinical nurse specialists at St Thomas Hospital. The purpose was to congratulate parents on the birth of their baby, reiterate why they had been contacted about the NBS, and provide details regarding what would happen next, including details of when

and where they needed to take their baby for confirmatory testing. It was also recommended that reliable condition-specific links to information sources be included. The text was drafted and revised with input from the co-design working group until they agreed that the language and style of communication were appropriate and all information for all 9 conditions currently screened for was included.

EBCD Process

The participants were asked to reflect and provide feedback on their experience of the EBCD process using the template provided by the EBCD toolkit [18]. This included a 5-point Likert-type scale ranging from *excellent* to *very poor*. All parents (21/21, 100%) rated viewing the composite film of parents' experiences as *excellent*, their experience of being filmed as *good* or *excellent*, meeting other parents and talking about their experiences as *excellent*, and the emotional mapping exercise as *good* or *excellent*. They felt that the priorities agreed upon at the end of the parent event reflected their own experiences of what needed to be improved. A total of 29% (5/17) of the health professionals provided feedback and indicated that their overall impression of the health professional feedback event was *excellent* and an *excellent* way to reflect on experiences at work.

Discussion

Uncertainty has been described as the single common challenge faced by patients who receive health care and the health professionals who provide it [37]. NBS by definition is not diagnostic and, as such, uncertainty in terms of clinical and prognostic outcomes is inevitable when communicating the initial NBS result [38]. In this study, parents and health professionals were able to successfully work together to identify priorities and develop co-designed interventions to improve the communication of positive NBS results using a modified EBCD approach.

Parents' Experiences of Receiving NBS Results

Consistent with previous research [9,10,13,39-42], parents in this study reported receiving NBS results in a range of ways, including face-to-face and via telephone and SMS text message, from a variety of clinicians, including nurses, physicians, and health visitors. The method used is, to some extent, influenced by the seriousness of the condition identified and the need for an immediate or less immediate response. MSUD and sickle cell carrier status would, for instance, be expected to be treated very differently in relation to the approach adopted. Furthermore, the content of the communication was less well defined and was, to some extent, determined by the person delivering the result. Current UK guidance states that the health professional delivering the news should be "appropriately trained" [43,44]. This is important as, similar to previous research [4,9,13,39,45], knowledge of the person communicating the result was considered important in this study to provide reassurance and allay parental fears.

In addition, parents in this study expressed the importance of the personal and professional attributes of the person delivering the news. In terms of personal attributes, this included being kind, empathetic, and supportive (physically and verbally) and possessing effective communication skills that allowed them to appropriately pace and tailor the information given and take the necessary time to explain the condition and answer parental questions. In terms of professional attributes, this included being perceived as a specialist, being credible, and working in an organization recognized as a center of excellence. The importance placed on knowledge and attributes of the person

communicating the positive NBS result to families provides further support for the widespread use of specialist screening nurses who not only have knowledge of all conditions included in NBS but have also undergone relevant training related to breaking bad news and possibly even have counseling skills.

As previously reported [13,39], positive NBS results were associated with negative parental reactions, including feeling nausea, shock, disbelief, fear, and sadness. Previous research has reported the impact on parents [4,6,11] as well as on parent and child relationships [8] and family relationships [46,47]. This was reflected in the results of this study as parents talked about the impact on their relationship with the affected child, including being scared to bond with their child and the fear of being overprotective. In this study, the impact of the diagnosis on parental relationships ranged from bringing them closer together to causing a strain on the parental relationship. Parents also talked about the impact of sharing the news with family and friends; associated with this were feelings of responsibility, guilt, and a lack of understanding.

Health Professionals' Experiences of Delivering NBS Results

The experiences of health professionals delivering positive NBS results have been published elsewhere [1]. In summary, health professionals invested a lot of time and energy ensuring that the communication of positive NBS results to families was parent- and family-centered, but this could be influenced by the challenges they experienced, including inadequate information on the NBS card and parental reactions. As mentioned, a variety of methods for the delivery of positive NBS results have been reported previously [9,10,13,39-42] that are often determined by the seriousness of the condition. In this study, it became apparent that this was also to some extent dependent on local arrangements. The COVID-19 pandemic meant that telemedicine rapidly and unexpectedly became the medium for health consultations that had previously taken place face-to-face. Other research has indicated that staff found the use of telemedicine for the delivery of NBS results during the COVID-19 pandemic safe and effective [48], and recipients also considered it an acceptable alternative to face-to-face communication. Therefore, going forward, this may be an acceptable means of delivering positive NBS results to families that could be time-saving and, therefore, cost-effective if the content is well considered and the person delivering the result is knowledgeable about the relevant condition.

In addition to parental experiences, this study furthers our understanding of health professionals' experiences with communicating positive NBS results to families. Health professionals involved in communicating positive NBS results are passionate about making sure that, although the message is distressing for parents, it is communicated well. Variations in communication practices continue to exist and are influenced by many factors, including the resources available and the lack of clear guidance. This affected not only the methods used to communicate positive NBS results but also the content of the communication to parents. This is supported by previous research conducted both nationally and internationally [4,6,41,49] suggesting that further guidance may be needed to

ensure a more cohesive approach that meets the needs of parents and health professionals while being sensitive to the subtleties of each condition. However, the issue of finite resources and the need to prioritize them also requires careful consideration. Nevertheless, with clear evidence of the deleterious effects of poor communication practices on parents [4-12], this variability is neither reasonable nor conducive to building a positive rapport with families. This is vital to ensure concordance with treatment regimens and trust in health professionals to maximize outcomes for the children.

Co-designed Interventions

To respond to the experiences and issues raised by parents and health professionals, EBCD, an established technique for gathering experiences and for co-design, was used [20-22,27,32,50-52]. It has been applied for the first time in this study to explore parents' and health professionals' experiences with the communication of positive NBS results. The process has enabled the prioritization of stakeholder requirements and the identification of co-designed solutions and additions to existing processes.

The co-designed interventions (changes to the NBS card; condition-specific, standardized laboratory proformas; condition-specific communication checklists; and an email or letter template to provide information to families following the communication of a positive NBS result) tackled different stages of the screening journey and areas where the participants felt that communication could be improved to minimize the anxiety and uncertainty experienced. These tools have been tailored to guide health professional communication with the aim of providing a more consistent experience. The interventions have subsequently been piloted at 2 sites; findings from this have been published elsewhere [15].

EBCD can be time-consuming and logistically challenging [27]; modifying the process has been shown to reduce costs [27]. The *Rethinking Strategies for Positive Newborn Bloodspot Screening Result Delivery: a process evaluation of co-designed interventions* project has been delivered during the COVID-19 pandemic; this has presented challenges in terms of bringing parents and health professionals together, a challenge that may continue for some time worldwide. We have adapted to these circumstances by using Basecamp as a collaborative tool enabling web-based EBCD outside the health care setting.

Strengths and Limitations

This is the first known study that has explored communication pathways for positive NBS results from the laboratory to parents via clinical teams. Health professionals were recruited from clinical teams involved in managing all the conditions currently included in the NBS program. This increases the transferability of the study findings as previous work has mainly focused on CF and SCD. This is the only known study that has used EBCD to bring stakeholders together to develop co-designed interventions to improve the communication of positive NBS results.

In terms of limitations, health professionals were recruited via email; those with a pre-existing interest in this topic may have been more likely to self-select into the study. They may

communicate results differently from providers who did not participate in the study, which may have biased the findings. However, health professionals were recruited from clinical teams involved in managing all the conditions currently included in the NBS program, which could have contributed to both the depth and breadth of the data collected. The researchers are experienced in this field, which may have biased data collection and analysis. Most parent participants were White British, which may limit the transferability of the findings.

Implementation and Further Research

COVID-19 has meant that web-based consultations via platforms such as Microsoft Teams and Zoom are being used to communicate with families about their children's positive NBS results. These have been described as an approximation to face-to-face interaction and are considered a visual upgrade of telephone consultations [53]. Initial studies that have explored these as a means of communicating positive NBS results to families suggest that they could be a safe and effective method for the delivery of positive NBS results to families [15,48]. Evidence suggests that video consultations (often referred to as telemedicine) have been viewed more favorably than telephone consultations [54]. The benefits of building rapport before using web-based approaches were found during teleconsultations in primary care during the lockdown [55]. The opportunities for using these web-based methods in NBS require further exploration to ensure that they are used appropriately, that the content of the message continues to be carefully crafted, and that the people involved are knowledgeable about the specific condition. However, a hybrid approach could act as a potential solution to address parental preferences, in particular face-to-face communication with their significant other present, communication via a condition-specific expert, and the clinical need for the timely provision of results.

In addition to the delivery of health care remotely, the pandemic has required web-based research and development. The adaptation of EBCD to include web-based methods could reduce costs while being easier to schedule. Adopting a web-based approach also has the potential to mitigate the imbalance of perceived power hierarchies [34] when patients and health professionals work together or, conversely, make it challenging to build a rapport. In this study, we benefited from the early stages of the process being run face-to-face, enabling relationships to develop. It is likely that a blended approach including face-to-face and web-based methods would help build effective relationships while offering flexibility and adaptation to the needs of parents (eg, childcare needs) and health professionals (eg, busy schedules). We argue that, as hybrid or blended ways of working are of increasing focus, the consideration and evaluation of different models of delivery for application in health care design would be beneficial.

Conclusions

Staff involved in communicating positive NBS results are passionate about making sure that, although the message is distressing for parents, it is communicated well. Despite this, variations in communication practices continue to exist. This is influenced by many factors, including the resources available and the current lack of clear guidance. Parents and health

professionals were able to successfully work together to share experiences, identify priorities, and develop potential solutions to improve the communication of positive NBS results to parents. The resulting co-designed interventions address communication at different stages of the communication

pathway to improve the experiences of parents receiving positive NBS results for their children. Adopting a hybrid approach to EBCD that incorporates web-based co-design working groups could enhance the success of future EBCD projects.

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Conflicts of Interest

None declared.

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Abbreviations

CF: cystic fibrosis
EBCD: experience-based co-design
FST: family systems theory
MSUD: maple syrup urine disease
NBS: newborn bloodspot screening
NHS: National Health Service
PPI: Patient and Public Involvement
SCD: sickle cell disease

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Original Paper

Meeting the Burden of Self-management: Qualitative Study Investigating the Empowering Behaviors of Patients and Informal Caregivers

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Abstract

Background: Patient empowerment is an important concept and a movement toward person-centered care of patients with chronic conditions. Nevertheless, to date, most research on empowered patients or informal caregivers has been conducted from a narrow clinical perspective. Such research has mainly focused on how health care professionals can empower patients to increase self-care or compliance with treatment. Research on empowered patient and informal caregiver needs and self-empowering activities is scarce.

Objective: We aimed to explore empowering behaviors from a patient and informal caregiver perspective in the context of self-management and to understand how health care can support such behaviors better.

Methods: We used an exploratory, qualitative study design. A total of 15 semistructured interviews and 6 focus group interviews were conducted with 48 patients and informal caregivers. We analyzed the interviews using thematic analysis and used a directed content analysis to analyze the focus group interviews.

Results: A total of 14 patterns of empowering behaviors were identified that were characterized by several exploratory and influencing activities performed by the participants. The participants expressed a desire to be more active in their care than what is expected and supported by health care professionals. The participants also desired better support for activities imposed on them by health care professionals.

Conclusions: To enable a transformation of the health care system to better support self-empowering behaviors, there is a need to develop self-management approaches from a patient and informal caregiver perspective.

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KEYWORDS

behaviors; chronic conditions; model of illness-related work; empowerment; self-management

Introduction

Background

The academic discourse increasingly maintains the view of the patient as an empowered and knowledgeable participant leading the way for their peers in a difficult health care setting. The notion of the empowered patient is further conceptualized in different concepts in the literature. Ferguson and Frydman [1] wrote about the concept *e-patient* in the early 2000s, inspired by the digital development within society that was reflected in many patient behaviors [1]. *e-Patient* describes patients or informal caregivers (such as a family members or other persons with a close relationship to the patient) who use the internet to find health-related information and web-based peer communities. *e-Patients* are further described as “engaged, enabled, equipped and empowered” in relation to their health or in collaboration with health care professionals [2]. *e-Patients* have also been shown to generate their own health data which they learn from and share, as well as create innovative solutions from [3]. Furthermore, the Department of Health and Social Care in the United Kingdom introduced the concept of *expert patients*, to describe user-led self-management for chronic conditions [4]. Expert patients take the role of mentors to other patients, with their significant knowledge and skills in self-management and patient participation. Expert patients are recognized to be valuable in clinical situations, research, representing patients in committees, or lobbying to health care authorities [4,5]. Both concepts are related to the concept of *lead user* in the field of innovation research. Lead user theories describe individuals who create innovative solutions to meet their own specific needs and predict needs for the consumer population in general [6]. Notably, patient innovations can sometimes enable better coping strategies and improved self-management overall [7].

Although the general direction in research and practice increasingly acknowledges patients’ abilities and potential as meaningful partners in different domains of health care, we noted 2 important shortcomings in the literature [8-11]. The first shortcoming is that most research on patients’ or informal caregivers’ empowering behaviors has been conducted from the perspective of health care professionals. Such research primarily focuses on how patients and informal caregivers can contribute to health care and how they fit into the needs of the health care system [8-11], rather than what these individuals need from health care. According to Zimmerman [12], patient empowerment consists of 3 components: the belief in one’s own capability to influence the situation (intrapersonal component), the understanding of which actions to take to achieve a desired outcome (interactional component) and engaging in specific types of behaviors to exercise control and influence (behavioral component) [12]. However, Eskildsen et al [13] state that patients can only become empowered if given the opportunity by health care professionals [13]. Thereby, drawing upon the definition of empowerment as a relational concept [14] is dependent on health care professionals conveying power to a homogeneous group of patients [15]. This further emphasizes the health care professionals’ perspective. Studies on self-empowering aspects of patient empowerment are largely

neglected in that perspective, and those aspects could be extended within the model of illness-related work describing patient self-management [16]. The second shortcoming we identified is that although the skills of empowered patients and informal caregivers have been described [2,17,18], there is still a lack of structured patterns of behaviors and the factors that influence them. This knowledge gap and the lack of a thorough examination of patients’ and informal caregivers’ needs, and expectations are addressed in this study. The aim of this study is to explore empowering behaviors from a patient and informal caregiver perspective in the context of self-management and to understand how health care can support such behaviors better.

The Model of Illness-Related Work as Theoretical Background

The model of illness-related work by Corbin and Strauss [19] describes medical management, role management, and emotional management as tasks for gaining greater control when performing self-management [16,19]. The model lists 6 self-management skills: problem solving, decision-making, finding and utilizing resources, patient–health care professional collaboration, action planning, and adapting skills regarding one’s condition [9,20]. The model provides direction for nurses to practice and teach self-management strategies [16,19]. The model’s 3 self-management tasks provide a description of healthy and interventional behaviors. Medical management includes taking recommended medication, following directives for hygiene before, for example, surgery, or using assistive devices or tools to manage a disability. Role management describes how patients need to maintain or create new role-specific behaviors in line with their chronic condition. This can include navigating through the health care organization, finding new ways to perform physical activities, or finding correct information about their condition. Emotional management entails dealing with emotional aspects of having a lifelong condition, such as coping, depression, grief, and existential beliefs [16].

The Taxonomy of Burden of Treatment as Theoretical Background

The 3 tasks presented in the model of illness-related work illustrate the complexity of managing a chronic condition. This complexity is further explained from a patient’s perspective by the taxonomy of burden of treatment [21]. To construct this taxonomy, patients with chronic conditions were asked to recount the structural burden they had to handle every day. These burdens included the need to coordinate between health care professionals, manage personal and economic factors owing to their condition, perform lifestyle changes, find information, and learn about their condition and create relationships (Multimedia Appendix 1 [1]). These burdens are described as being imposed on patients as they perform self-management and could lead to struggle with adherence to treatment and care, as well as poor quality of life [21,22].

Methods

Overview

This exploratory study followed a qualitative approach in 2 consecutive stages. The first stage consisted of semistructured interviews with 15 patients with chronic conditions or informal caregivers. All participants described themselves as highly empowered regarding their self-management and in collaboration with health care. The second stage consisted of 6 focus group interviews with a broader group of 33 patients with chronic conditions or informal caregivers. All participants were from different parts of Sweden. A total of 9 interviews were conducted via telephone and 6 via face-to-face interviews. All focus groups were performed physically in settings close to participants' homes. The semistructured interviews were analyzed using thematic analysis and the resulting categories were used as key concepts to guide a directed content analysis of the focus group data.

Recruitment and Sampling

Participants in both stages were recruited using purposive sampling [23,24]. For the semistructured interviews, participant recruitment was conducted across Sweden through a web-based announcement on a webpage from a project called "Patient Lead Users," which addressed people with chronic conditions or their informal caregivers nationally. The announcement included a request for empowered patients and informal caregivers to nominate themselves or someone else as being actively engaged in collaboration with health care as well as self-management. Further inclusion criteria were age >18 years and having experienced ≥ 1 chronic condition. Of the 67 self-nominated or suggested participants, 10 (15%) patients and 5 (7%) informal caregivers were selected by the authors to cover different ways of being active within their self-management, as well as different chronic conditions, sex, age, and geographic locations. The study sample for stage 2 consisted of 33 participants distributed over 6 focus groups. In this stage, patients with chronic conditions and informal caregivers were approached and screened through patient associations or through employed peer support workers within different geographic regions in Sweden. This was done after analyzing stage 1. The inclusion criteria were aged > 18 years and had chronic conditions.

Content Development

The semistructured interviews in stage 1 consisted of open-ended questions covering 4 themes: background, your health journey, health behaviors, and your role (Multimedia Appendix 2 [2]). The chosen themes were based on identified knowledge gaps in the literature; the lack of knowledge about patients' and informal caregivers' needs in relation to their health journey, how they act (health behaviors), and what influences their behaviors (your role). A total of 5 pilot interviews were conducted to test the questions provided in the interview guide. Data from the pilot interviews were not

included in the study results. In stage 2, a multiple-category design was used with different types of participants and chronic conditions [25]. The questions in the protocol for the focus groups (semistructured interview guide, Multimedia Appendix 3 [3]) were developed from the analysis and results of the semistructured interviews conducted in stage 1.

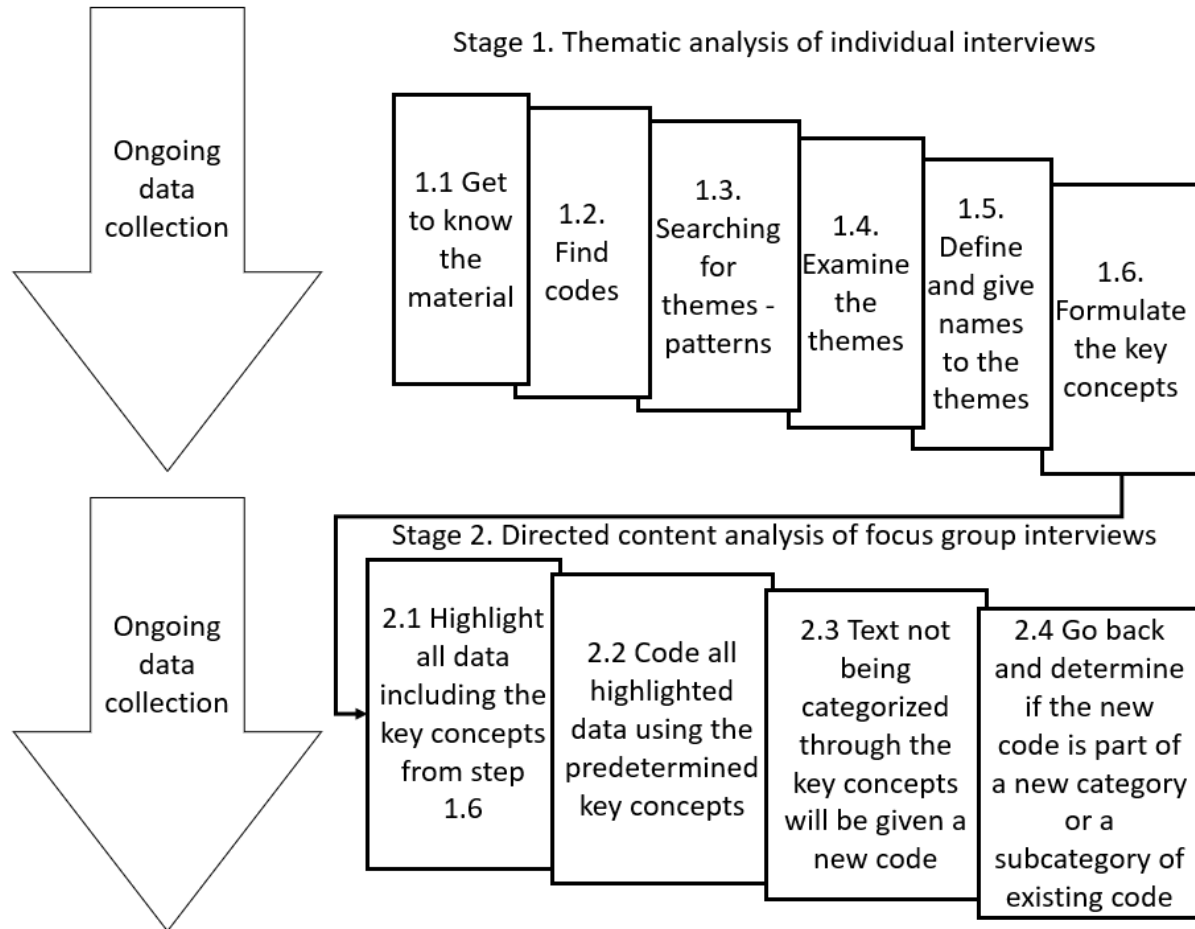
Data Collection

Data for both stages were collected by the first author and 4 coworkers from the Patient Lead User project between November 2017 and September 2019. The interviews consisted of 6 face-to-face interviews at a location convenient for the participants and 9 interviews over telephone when face-to-face interviews were not possible because of their condition or long distances. The interviews lasted an average of 44 minutes, with a total duration of 656 minutes, and a SD of 7.4. In stage 2, each focus group consisted of a moderator and 1 or 2 observers. The sessions lasted for an average of 103 minutes, with a total duration of 618 minutes, and a SD of 13.2. All focus groups were performed physically in settings close to participants' homes. The semistructured interviews and focus group sessions were recorded and transcribed verbatim. Transcribed data were returned to those participants who requested it and provided feedback on the findings when necessary. Saturation was reached [24] after 12 interviews in the first stage and after 5 focus groups in the second stage. To verify the results, 3 additional semistructured interviews and 1 additional focus group were conducted. No further recruitment was necessary in addition to the original sample.

Data Analysis

In stage 1, a thematic analysis was performed by all authors in parallel with the data collection. This is a flexible and inductive approach to analyze the data for the semistructured interviews [26,27]. Six phases were included: (1) familiarization to get acquainted with the data, (2) categorization of the data into units according to how the meaning of the data shifted, (3) finding patterns between the units to create themes, (4) situating all coded data into themes, (5) naming the themes according to their essence of how they fit into the aim of the study, and (6) formulating the key concepts from the categories within the themes [26]. The first stage resulted in 11 categories. These categories were used in stage 2 to further test and validate the knowledge from the semistructured interviews (Figure 1). Directed content analysis was used for data analysis [28,29], and the categories from stage 1 were used as key concepts to initiate the coding process. The authors followed 4 steps to examine how the categories emerged as behaviors related to self-management aspects [29]. (1) All data from the focus groups were coded and, when applicable, mapped into key concepts from the thematic analysis. (2) Subcategories were developed. (3) Data not matching one of the key concepts were assigned a new code, and (4) 3 new exploratory behaviors were established as categories.

Figure 1. Illustration of the analysis for the whole study.



Ethics Approval

Ethics approval (decision 2015/1572-31/4 for interviews and 2018/2294-32 for focus groups) was provided by the Stockholm Regional Ethical Review Board. Written information about the purpose of the study, management of the data, and the option to opt out at any time was provided to the participants before the interviews and focus groups started. All participants signed informed consent forms after receiving oral and written information.

Results

Overview

Participant characteristics included age, occupation during the time of interviews, years since diagnosis, sex, and different chronic conditions (Table 1 and Textbox 1). The analysis of the interviews resulted in 11 categories: *self-care expert, academic, patient researcher, tracker, innovator, entrepreneur, communicator, mentor, health care coordinator, health care*

partner, and activist (see the white boxes in Figure 2). From the focus groups, 3 new exploratory behaviors were elicited: *knowledge seeker, coping expert, and exposed* (see gray boxes in Figure 2). The findings showed 2 major classes of empowering behaviors related to participants’ self-management activities: *exploratory* and *influencing behaviors*. These were illustrated as 2 overarching themes, where theme 1 described patterns of the exploratory phase of participants’ self-management, gaining experience, and knowledge of their condition. Theme 2 described patterns of the influencing part of self-management when the participants wanted to share their lived experience and knowledge with their peers and at the health care system level (Figure 2).

The participants adopted several patterns of behavior from both the themes, albeit often as a stepwise approach that spanned over several years and in different contexts. However, several of the exploratory patterns of behaviors were kept in parallel with the influencing patterns of behaviors, such as the self-care expert, knowledge seekers, academics, patient researchers, and trackers.

Table 1. Characteristics of participants (N=48).

Participant characteristic	Value, n (%)	
	Stage 1	Stage 2
Age (years)		
18-45	5 (33)	8 (24)
46-65	7 (47)	14 (43)
>66	3 (20)	11 (33)
Occupation		
Retired	4 (27)	15 (46)
Sick leave	2 (13)	4 (12)
Working	9 (60)	11 (33)
Studying	N/A ^a	3 (9)
Years since diagnosis		
<5	3 (20)	8 (24)
6-10	7 (47)	13 (40)
>10	5 (33)	12 (36)
Sex		
Female	10 (67)	25 (76)
Male	5 (33)	8 (24)
Participant type		
Patient	10 (67)	27 (82)
Informal caregiver	5 (33)	4 (12)
Both	N/A	2 (6)

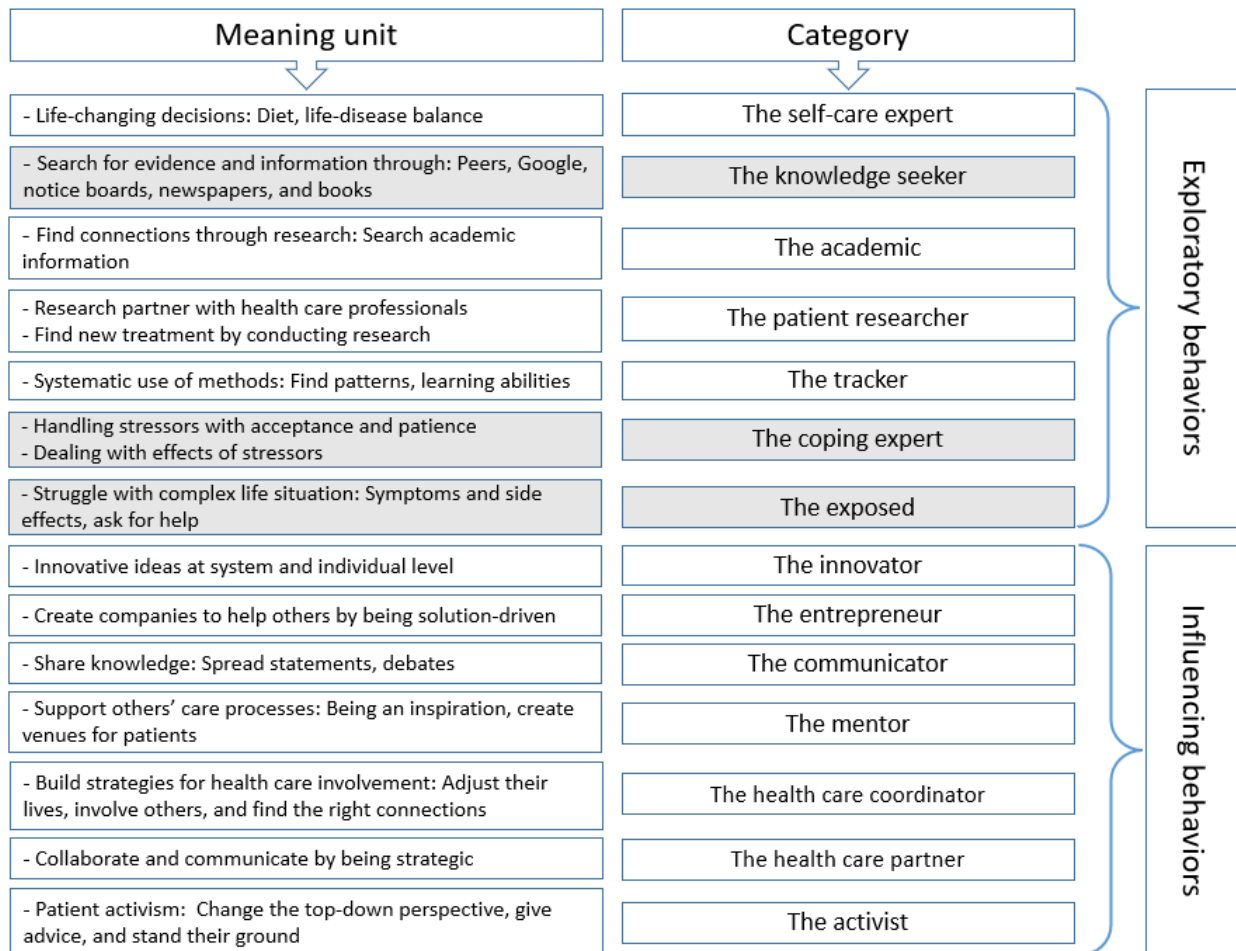
^aN/A: not applicable.

Textbox 1. Chronic conditions presented by the participants.

Chronic condition

- Brain neoplasms
- Breast neoplasms
- Colonic neoplasms
- Connective tissue disease
- Cyst-liver and Cyst-kidney
- Diabetes type 1 and 2
- Down syndrome
- Fatigue syndrome
- Fibromyalgia
- Heart condition
- Hypersensitivity
- Irritable bowel syndrome
- Kidney failure
- Kidney neoplasms
- Liver neoplasms
- Meningocele
- Mental illness
- Motility disorder
- Multiple sclerosis
- Myalgic encephalomyelitis
- Myelodysplastic syndrome
- Myocardial infarction
- Ovarian neoplasms
- Parkinson disease
- Prostatic neoplasms
- Pulmonary fibrosis
- Rheumatic disease
- Stroke
- Systemic sclerosis
- Thymus neoplasms
- Uterine neoplasms
- Whiplash injury

Figure 2. Illustration of key concepts and themes.



Patterns of Exploratory Behaviors

Patterns of exploratory behaviors entail how participants contemplated their own situation and explored new ways to improve their situation. We identified the following patterns of behaviors: *self-care experts, knowledge seekers, academics, patient researchers, trackers, coping experts, and being exposed.*

By developing self-care strategies, making far-reaching lifestyle changes, and making life-changing decisions to create a balance in life, the *self-care expert* included patterns of behaviors found in several participants:

My clinician suggested dialysis for my kidney failure. However, I have Googled and knew it would give me 4-5 years to live, so I refused. I wished to get a transplant, if I still was alive after 5 years because of my kidney neoplasms. So, I asked my physician what I can do in my self-management to achieve that goal. [Patient from stage 1, male]

Seeking knowledge from the literature, social media, or other digital sources is a major part of participants' self-management. This provides for learning opportunities as a *knowledge seeker* or to perform a more systematic search of available literature, compiling research, finding connections to their own condition, and stay updated on the latest scientific articles and evidence as an *academic*. These proactive actions allow participants to

find new ways of managing their condition and are often performed when the information from health care is insufficient:

The physician rarely talks about the future of my Parkinson disease. So, I push a bit because I have found information about something I want to test. Then I think about those who are not as well informed, do they not get the same care as I do? [Patient from stage 2, female]

Suffering from two heart attacks, I decided it would not happen to me again. I started to read the literature of preventive measures, but it was too generic. So, I did my own review of scientific articles, to find the triggering aspect of my disease... [Patient from stage 1, male]

Sometimes, an academic or knowledge seeker transforms into a *patient researcher* when developing a partnership with health care professionals to examine research to identify potential new treatment, use scientific methods to investigate their health issues, and engage as patient partners in research programs:

I found studies suggesting a biological medication for my systemic sclerosis. However, the medication was not yet approved. But with a great relationship and exchange concerning research, my physician helped by motivating a prescription for that medication... [Patient from stage 1, female]

My therapist and I are “research friends.” Together we try to tackle new aspects of my mental illness... I think it is interesting since she does not try to be superior to me, even though she has a lot of knowledge... but we are on the same page regarding how to perform research together... [Patient from stage 2, female]

The *tracker* includes patterns of behaviors found in several participants by systematically using methods to measure different health-related aspects (such as sleep, mood, food, physical activity, etc). The participants used digital technologies, paper and pen, or their mind to establish patterns, and learn through data to modify treatment or other health-promoting activities. This would be done to achieve deeper learning, better health, or better communication with health care professionals:

I adjusted time and dosage during the day, not exceeding my daily maximum dosage set by the physician. It resulted in me improving my health... and my self-efficacy regarding health care collaboration increased, since I realized that physicians can only give me guidelines regarding my Parkinson disease, then it is up to me to adjust according to my situation. [Patient from stage 1, female]

Depending on how I feel and what feels relevant for me and my Multiple sclerosis, I perform self-tracking... It is related to food intake, and I have some classifications of how I measure health status. Then I also optimize my physical activity so I do not sit still all day and then believe I could compensate that with an hour at the gym. This is done with my smart watch, reminding me to move every hour... [Patient from stage 1, male]

To emotionally cope with their self-management, some participants became *coping experts* dealing with stressors in their everyday life, either by changing their emotional responses to different stressors, which could include delaying difficult activities, or by dealing with the stressor itself. Restoring energy through physical activities and working with acceptance are some techniques they used to reduce existing problems:

I am driving motorcycle, traveling, and taking long hikes, until I am too sick. Because I know that the day will come when I need those mental pictures to be able to cope and trying to stay alive with my growing cyst-kidney and liver. [Patient from stage 2, female]

I distanced myself from my Parkinson disease as long as possible... and I am quite happy since I am rather sensitive, so it was nice to be able to keep it all away from me. That I have not constantly thought of it for 11 years... instead I take it step by step. It has suited me well. [Patient from stage 2, female]

Some participants also experienced being *exposed*, trying to collaborate with health care professionals and struggling with a complex life situation in society and within their family. They described an emotional struggle, sometimes hiding from society, feeling lonely, and not belonging anywhere. However, being

exposed also included identifying problems that need to be solved:

The situation is complex since we [my husband and I] are both living with a chronic condition. I know how to live my life to manage my self-care for my Parkinson disease, but I'm not capable, since I need to take care of my husband with multiple diseases as well. But I cannot... leave him. I'm not there yet. [Patient and informal caregiver from stage 2, female]

I worry when walking in the city, because of my whiplash injury. I can get very dizzy just stepping off a curb. And perhaps the police might think I'm drunk [laughing]... [Patient from stage 2, female]

Patterns of Influencing Behaviors on the Individual and System Level

The patterns of influencing behavior are those that may change the surrounding environment. Such patterns of behavior were exemplified as follows: *innovator, entrepreneur, communicator, mentor, health care coordinator, health care partner, and activist*. Good ideas based on health and health care needs were often based on previous lived experiences of chronic conditions and knowledge from their working life.

Exemplifying patterns of behaviors for *innovators* are the needs of performing self-care and collaborating with health care in a better way or to help in a community of peers based on experiences of lacking information related to their specific situation. Coming up with novel solutions for their situation was often accomplished by using digital or other physical solutions:

I met two other patients who have multiple sclerosis and that never had been in contact with someone they could relate to... Then the idea was born to try to reach out to other young patients by programming a webpage for this target group, since we felt it was empowering to be able to talk to someone who really understands you. [Patient from stage 1, male]

Sometimes, *innovators* start organizations from their innovations, based on a strong need for more information or to help their peers. At other times, the participants become *entrepreneurs* based on their own health care experiences:

I was not given the correct treatment, which is very common for some diagnoses such as my genetic connective tissue disease, since there are no clear instructions for how to diagnose within primary care. The concept within the company is a process that has been digitalized and builds on trying to make it easier for primary care to refer you as a patient to the right specialist. If you do not get a referral to the right specialist, they do not know how to handle you. [Patient from stage 1, female]

I started a publishing company and wrote and published several books about my healthcare experiences as an informal caregiver to my wife with pulmonary fibroses and liver neoplasms. [Informal caregiver from stage 1, male]

Communicating with others about their lived experiences meant being a patient advocate, an inspiration, and making the disease visible. Several participants hoped to make a change for other peers in health care situations with their increased knowledge and acceptance in society, as well as using their professional knowledge. As a *communicator*, the participants could be working with companies, writing op-ed articles, using social media as a platform, or speaking at conferences in health care contexts:

Speaking at conferences or when writing article as an informal caregiver, there are two specific topics that I believe are connected: continuity and digitalization. To achieve a person-centered encounter, you need to combine that with the best suitable technology. I refuse to believe the choice is either to use technology or to have a physical encounter... [Informal caregiver from stage 1, female]

I believe it is appreciated when I inform healthcare professionals at meetings and conferences, about my everyday life with Diabetes type 1... I believe it could lead to better treatments if we could collaborate in a new way... The patient has so much valuable information that is needed in health care as well, and that is where my heart is, trying to bridge a gap between health care and the patients. [Patient from stage 2, female]

Whereas a *communicator* kept informing groups of people, others provide mentorship at an individual and personal level. By becoming role models for others through inspiration and paying it forward as a *mentor*, the participants made use of the knowledge they had acquired:

It is so rewarding to help my peers within Rheumatic disease... There are for example many people with foreign background in my region, and it is difficult for them to make themselves understood, and they might not ask for what they have the right to within society... I could work 100 percent just helping them, so they could improve their own chronic condition and health. [Patient from stage 1, female]

I need to travel far for my treatment for Prostatic Neoplasms. Luckily there are three of us always traveling together, since we have convinced health care that we need to have slots suiting all three of us. This mean a lot, especially for one of us who is all alone with no family member supporting him. Now he could go with us and get support and not feeling alone in this... [Patient from stage 2, male]

One major part of the participants' self-management was coordinating their care at different health care sites. This required considerable knowledge, developing special skills to manage different actors around the patient, building relationships, and finding pathways to the right resources in a solution-oriented way of thinking as a *health care coordinator*. Often, the patient's condition requires many health care contacts:

I need to coordinate primary care, the heart clinic and... the habilitation... as well as dental care since that is very important when having a heart failure. I

have tried to make them all collaborate... [Informal caregiver from stage 1, female]

Often, a deeper relationship and collaboration with health care professionals is crucial for the work as a health care coordinator. Some participants exemplified that their experience of a collaboration with health care professionals as *health care partners* is a great learning opportunity. Within these relationships, the participants also felt listened to and that health care professionals knew the participants had valuable information about their lives, for them to provide the best care. Even though increasing their collaboration with health care professionals was desirable, most of the participants experienced that it was difficult to achieve:

It has been challenging to represent my son in health care situations, since he is not good at explaining how he feels regarding his Down's syndrome and heart failure... and to make health care professionals listen and to understand our situation. Before I would yell and scream. Now, I've learned it is more effective to lower your tone, to be taken seriously. [Informal caregiver from stage 1, female]

I have been within health care with my rheumatic disease since I was 13 years old, and since it has been that many years an interest has grown within me to work with healthcare professionals and to become one myself. [Patient from stage 2, female]

A few participants had the urge to change the health care system as they did not believe it was person-centered. They challenged the paternalistic structures trying to make a change in policies and structures related to their health care needs and health. These *activists* were acting as influencers on the web and offline. This was to help themselves and other peers stand their ground:

It is important to connect with people who are already interested in the topic, to be able to generate a change. There is no use banging one's head against a wall... You can always start with a small change, and not wait for the structure to change. I believe it is important with this bottom-up-perspective. [Informal caregiver from stage 1, male]

Discussion

Principal Findings

By exploring empowering behaviors in relation to self-management, we have identified different patterns that the participants follow. They are listed here with a descriptive term for each: *the self-care expert, the knowledge seeker, the academic, the patient researcher, the tracker, the coping expert, the exposed, the innovator, the entrepreneur, the communicator, the mentor, the health care coordinator, the health care partner, and the activist*. These patterns of behaviors are characterized by different activities that the participants perform for several years. None of the participants followed only one of these patterns but commonly adopted several patterns expanding over both exploratory and influencing behaviors. One example can be that a person starts with noticing a feeling of being exposed and vulnerable and addresses that by seeking knowledge and

building self-management experience. Furthermore, the participants used that experience to make a difference for others and change the health care system through activism and new innovations. This empowering process is characterized by learning experiences and adapting to the current situation, depending on previous knowledge and existing skills. A common theme in the participants' narratives, regardless of exploratory or influencing behaviors, was that they did not feel that they received support in their efforts from the health care system or society at large and sometimes even felt hindered when their behaviors were not in line with the expectations. The participants expressed having a desire to do much more than is currently expected by health care professionals, such as being a health care partner, an innovator, and a mentor. They also wanted better support for tasks that were not within their ability or interest but were imposed on them, such as acting as coordinators of their care.

With the current rather limited view on concepts such as self-management and patient empowerment within the existing literature and in the health care system, it is important to illustrate the concepts from a holistic perspective on patients and informal caregivers. This study contributes to the richness of empowering behaviors, illustrating how the participants extend their limits to influence the situation for themselves and for others. The participants know which actions to take within health care and in their self-care; however, the desired outcome is dependent on a functional collaboration with relevant actors. The study provides further evidence to the notion that patients and informal caregivers in some cases develop extraordinary behaviors and competencies that can serve as inspiration to others.

Findings in Relation to Theoretical Background

Our results on empowering behaviors extend on the model of illness-related work [16]. By applying a patient and informal caregiver perspective, we uncovered additional categories of work that need to be undertaken in the case of illness. These categories of work can primarily be categorized as part of role management, entailing digital activities, working to improve the health care system, collaborating with health care professionals, and finding ways to navigate the health care system. Moreover, role management could also mean that the participants found new solutions for their needs, started companies to meet the needs of their peers, searched for information outside health care, became mentors spreading inspiration and information to their peers, tracked symptoms, and used proactive behaviors such as being self-care experts to prevent further disease by doing more than expected. To cope with everyday life and the feeling of being exposed are included in emotional management. However, we do see that most behaviors are part of role management, having to engage in activities that were not part of their life before the illness. Medical management was not explicitly included in our participants' accounts, although we can see role management, such as the adjustment of medication based on tracking data being performed within the limits for medical management given by health care. This can be explained by the fact that the participants often had different views of what the most pressing tasks were rather than what was expected from the model of

illness-related work. Emphasis on medical management was also a central theme in the patient empowerment literature, where the focus was on disease management and health care interaction [15]. Similarly, the concept of patient participation is most often narrowly described from a health care perspective [30]. One important contribution of this study was that, in contrast to the previous literature, our findings emphasized the self-empowering aspects of patient empowerment.

The taxonomy of the burden of treatment helped us describe the participants' situation and understand where their behaviors arise from. The taxonomy of the burden of treatment describes how disease-related tasks are imposed on patients, how factors associated with these tasks are intensifying the burden, and how patients are affected by it [21]. In our results, the participants illustrated different situations of burden arising from personal circumstances and from the system level. The participants experienced everyday life burdens living with a chronic condition, such as dealing with stressors and feeling exposed within the society and family. Burdens could also entail a lack of collaboration with health care professionals, including insufficient information, not to be taken seriously, and being misdiagnosed. When it comes to coordinating health care at a system level, the participants illustrated challenging situations trying to navigate for themselves or for their next of kin within different health care situations. These burdens correspond well with the described aspects in the taxonomy [21], illustrating the burdens of lifestyle changes, nonworking collaboration with health care professionals, understanding of their condition and treatment, and emotional aspects. The empowering behaviors resulting from our participants' narratives appeared as a paradoxical driving force toward increased autonomy and empowerment, moving forward from these obstacles in life. The participants understood that it was up to them to make changes within their self-care as health care professionals could only give them guidelines and not specific instructions to gain better well-being and health. By becoming mentors, communicators, and activists, the participants worked for change within the health care system. The participants shared what they learned through their lived experience and pursued a mutual learning experience with health care professionals.

Strengths of Using Two Different Approaches of Data Collection

Our 2 different approaches to data collection gave us the opportunity to first gain an inductive and deep knowledge and thereafter to test the key concepts in 6 focus groups using an abductive approach. This also provided us with in-depth data when participants were inspired by each other and considered different aspects of their behaviors and activities than they would in single interviews. Including a larger group of participants mitigated the risk of self-management activities being performed only by powerful patients and informal caregivers with capital [31]. Self-management was performed by all participants; however, behaviors might differ depending on being an e-patient, expert patient, or lead user, or if the participant belonged to a late majority when it came to behavioral change. This was a process of reducing uncertainty regarding self-management behavior [32]. Behaviors performed by these early adopters are important for the development of

self-management approaches, since the late majority of patients and informal caregivers tend to follow later on and make use of these solutions for better health [33].

Limitations and Further Research

This study did not seek to perform a personality categorization of behaviors but instead illustrates different types of empowering behaviors as described by the participants. The sample was based solely on participants who were able to manage their or a family member's condition. The consolidated criterion for reporting qualitative research—COREQ checklist—was used to ensure the reliability of the data and maintain transparency throughout the study (Multimedia Appendix 4) [34,35]. In addition, having empowering behaviors does not solve the power inequality in a paternalistic context [36]. Therefore, future research needs to consider the different types of behaviors from a health care perspective to explore how these behaviors are received by health care professionals and their rather limited understanding of patients' self-management. This is important to increase the support from health care professionals.

Conclusions

Keeping a strict patient and informal caregiver perspective, this study provides an in-depth understanding of the participants'

empowering behaviors and emphasizes the richness of self-empowering aspects of patient empowerment by extending on the model of illness-related work. This notion enables a perspective of what the participants can and want to do within their self-management and in collaboration with health care. The result illustrates how the participants extend their limits to influence the situation for themselves as well as for others in various ways and as a paradoxical driving force moving away from the obstacles illustrated by their everyday life stories, as well as described in the taxonomy of the burden of treatment. Today, patients and informal caregivers are part of a system that is not based on their needs; yet they are the main users. However, their behavior might differ depending on whether they are early adopters or late majorities when it comes to behavioral changes. Behaviors performed by early adopters are important for the development of self-management approaches as peers tend to follow later and make use of these solutions. To enable a transformation of the health care system to support patient empowerment and empowering behaviors, there is a need to develop solutions from a user perspective. This will increase the use of patient self-management and improve health care toward a more person-centric system.

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Authors' Contributions

All 5 authors contributed equally according to the International Committee of Medical Journal Editors [37]. All authors designed the study, analyzed the data, and drafted the manuscript. The first author (PhD student) collected the data and transcribed all the interviews with an external company. All authors have experience and training in qualitative research. This manuscript version was approved by all authors.

Conflicts of Interest

None declared.

Multimedia Appendix 1

The taxonomy of burden of treatment.

[PNG File , 212 KB - [jopm_v14i1e39174_app1.png](#)]

Multimedia Appendix 2

Interview guide: stage 1.

[DOC File , 26 KB - [jopm_v14i1e39174_app2.doc](#)]

Multimedia Appendix 3

The protocol for focus groups: stage 2.

[DOC File , 27 KB - [jopm_v14i1e39174_app3.doc](#)]

Multimedia Appendix 4

COREQ (consolidated criterion for reporting qualitative research) checklist.

[PDF File (Adobe PDF File), 532 KB - [jopm_v14i1e39174_app4.pdf](#)]

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Original Paper

Text Messages to Support Caregivers in a Health Care System: Development and Pilot and National Rollout Evaluation

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Abstract

Background: Although there are many interventions to support caregivers, SMS text messaging has not been used widely.

Objective: In this paper, we aimed to describe development of the Department of Veterans Affairs (VA) Annie Stress Management SMS text messaging protocol for caregivers of veterans, its pilot test, and subsequent national rollout.

Methods: The stress management protocol was developed with text messages focusing on education, motivation, and stress-alleviating activities based on the Resources for Enhancing All Caregivers Health (REACH) VA caregiver intervention. This protocol was then tested in a pilot study. On the basis of the pilot study results, a national rollout of the protocol was executed and evaluated. Caregivers were referred from VA facilities nationally for the pilot and national rollout. Pilot caregivers were interviewed by telephone; national rollout caregivers were sent a web-based evaluation link at 6 months. For both evaluations, questions were scored on a Likert scale ranging from completely disagree to completely agree. For both the pilot and national rollout, quantitative data were analyzed with frequencies and means; themes were identified from open-ended qualitative responses.

Results: Of the 22 caregivers in the pilot study, 18 (82%) provided follow-up data. On a 5-point scale, they reported text messages had been useful in managing stress (mean score 3.8, SD 1.1), helping them take care of themselves (mean score 3.7, SD 1.3), and making them feel cared for (mean score 4.1, SD 1.7). Texts were easy to read (mean score 4.5, SD 1.2), did not come at awkward times (mean score 2.2, SD 1.4), were not confusing (mean score 1.1, SD 0.2), and did not cause problems in responding (mean score 1.9, 1.1); however, 83% (15/18) of caregivers did not want to request an activity when stressed. Consequently, the national protocol did not require caregivers to respond. In the national rollout, 22.17% (781/3522) of the eligible caregivers answered the web-based survey and reported that the messages had been useful in managing stress (mean score 4.3, SD 0.8), helping them take care of themselves (mean score 4.3, SD 0.8) and loved ones (mean score 4.2, SD 0.8), and making them feel cared for (mean score 4.5, SD 0.8). Almost two-thirds (509/778, 65.4%) of the participants tried all or most of the strategies. A total of 5 themes were identified. The messages were appreciated, helped with self-care, and made them feel less alone, looking on Annie as a friend. The caregivers reported that the messages were on target and came when they were most needed and did not want them to stop. This success has led to four additional caregiver texting protocols: bereavement, dementia behaviors and stress management, (posttraumatic stress disorder) PTSD behaviors, and taking care of you, with 7274 caregivers enrolled as of February 2022.

Conclusions: Caregivers reported the messages made them feel cared for and more confident. SMS text messaging, which is incorporated into clinical settings and health care systems, may represent a low-cost way to provide useful and meaningful support to caregivers.

KEYWORDS

mobile health; mHealth; self-care; veterans; family caregivers; emotional stress; burden of illness; self-efficacy; mobile phone

Introduction

Background

Caring for a loved one can substantially impact the emotional and physical well-being of the caregiver. Caregivers report higher levels of psychological distress, depressive symptoms, anxiety, stress, and emotional difficulties than the general population. They also report lower self-ratings of physical health, higher rates of chronic disease, and fewer self-care behaviors [1]. Social support, education, and skills training, for example, coping skills, problem-solving, cognitive reframing, and stress management, have all been shown to be beneficial for caregivers [1]. Multicomponent interventions that can be targeted to caregivers' specific concerns combining these strategies have generally provided stronger evidence of positive benefit for caregivers [2,3] in the form of reduced depressive symptoms, anxiety, and burden [1,4]. To support caregivers, researchers have tested interventions that vary across format (eg, face-to-face, telephone, inclusion of audio or video, printed materials, and websites), content (single focus or multicomponent), and length (1-48 sessions). Caregiver interventions are generally synchronous and delivered either in-person or by telephone or telehealth [2,5].

Caregivers have been shown to be receptive to technology if the technology is compatible with values and abilities and is tailored toward the caregiver [6]. Reviews of web-based interventions for caregivers of older persons and of persons with dementia show that support by a peer group [7-9], contact with a provider [7,10], assistance with decision-making and problem-solving [7,10], support [7-10], and information [7-10] included as part of a multicomponent intervention have the most positive outcomes. Mobile apps for caregivers also provide them ways to learn [11,12], interact with clinical experts [12], take care of themselves [11,12], and manage the care of their care recipient [12].

There is a need to expand family caregiver support in clinical practice and in diverse populations [3], but clinicians who deliver interventions may not always be available, and organizations may not be willing to invest in web-based interventions or mobile apps. SMS text messaging may be a simple way for health care organizations to address caregiver stress and their need for information. Texting in different formats, including synchronous or asynchronous chats, automated messaging, and push notifications, has been widely embraced as a health care intervention for a variety of conditions and purposes. Different types of text messages have been used, including reminders, information, supportive messages, and self-monitoring procedures. Multiple review articles have examined texting in health care in recent years, concluding that it can be beneficial clinically [13,14]. SMS text messaging improves patient engagement in treatment and outcomes [15,16] and adherence to treatment regimens and medications [17-20].

Despite their success in patient care, texting interventions have not been developed for caregivers [20] other than for parent caregivers of adolescents and children. Integrating SMS text messaging interventions more broadly into health care systems would help support patients, caregivers, and public health [21].

Text Messaging in the Department of Veterans Affairs

Automated Texting Platform for Veteran Self-Care, part of the Office of Connected Care of the Department of Veterans Affairs (VA), is an SMS capability that can send messages and interpret patient messages following a specified syntax to reply with preprogrammed (bidirectional) responses. Annie cannot decipher or respond without keywords. Veterans and caregivers can access the platform through their mobile phones or smartphones, and veterans also have access to an app. The platform is modeled after the United Kingdom's National Health Service's program Flo, after Florence Nightingale, the founder of modern nursing. Flo has been useful in symptom management, for example, in blood pressure control [22] and in early identification and management of complications after colorectal surgery [23]. However, two of the major benefits of Flo are education and feelings of support, control, companionship, and flexibility reported by patients [24]. The program of the VA is known as Annie, named after Lt Annie G Fox, Chief Nurse at Hickam Field, during the attack on Pearl Harbor on December 7, 1941, and the first woman to receive the Purple Heart for combat.

As of February 2022, Annie had 43,229 current users who were registered and had the capability to be on a protocol and 363 different protocols focusing on education and self-care, including disease-specific reminders (eg, diabetes foot care), behavior change encouragement and motivation (eg, exercise and weight loss prompts and breathing and relaxation strategies), medication and symptom monitoring (eg, blood pressure), and treatment adherence reminders and monitoring. Veterans using Annie receive motivational or educational messages that do not require a response or automated prompts to track and monitor their own health. These prompts only allow preprogrammed responses, such as typing in a word to receive further texts or reporting a blood pressure reading. Messages and patients' data are stored in the Annie system where clinicians can view the texts and readings as needed, but this is not a requirement for clinicians.

Annie, like Flo, not only has clinical benefits but also enhances the satisfaction and empowerment of the patients. Annie reminders have improved adherence to positive airway pressure for patients with traumatic brain injury and sleep apnea with subsequent improvement in sleepiness and cognition [25]. Patients with hepatitis C virus in Annie have shown not only improved adherence to medication regimens but also less distress at failing treatment [26]. Veterans who were receiving chemotherapy used Annie to report symptoms and seek further action if their symptoms warranted it. Patients felt empowered by being able to self-manage their symptoms, particularly when

their perceptions of doing well were validated [27]. Like Flo, Annie fosters a sense of connection and confidence in those who use it [28].

The sense of empowerment and companionship made Annie a perfect fit for caregivers. Although Annie had been successful with patients and there was enthusiasm for developing patient protocols, it had not been used for caregivers. The national Caregiver Support Program of the VA provided a receptive environment for providers and caregivers. A small caregiving texting pilot at the University of Tennessee Health Science Center showed that older African American caregivers were receptive to texting. Thus, in 2019, the VA Caregiver Center, part of VA’s Caregiver Support Program, began a collaboration with the VA Office of Connected Care to bring Annie SMS text messaging to caregivers of veterans. This paper describes the development of the Annie Stress Management texting protocol, its evaluation in a pilot study, and the evaluation of the subsequent national rollout into the broader VA system.

Text Messaging Caregiving Intervention (Pilot)

Methods (Pilot)

Overview

Caregivers of veterans enrolled in the Caregiver Support Program were eligible for the pilot study as these caregivers all had an electronic health record, a requirement for Annie registration. The pilot study was publicized to the national network of Caregiver Support Program teams of VA located at each facility. Caregiver Support Program staff from each VA facility sent a flyer about the study to each of their caregivers enrolled in the VA Caregiver Support Program. If a caregiver was interested in participating, staff registered the caregiver in Annie and then sent the caregiver’s information to the Caregiver Center Annie coordinator. The Annie coordinator then reached out to the caregiver and completed a short screening interview

to ensure that the caregivers could receive text messages and were willing to participate. Once a caregiver agreed to participate, the Annie coordinator mailed consent forms and scheduled a time to go over them over the phone. Caregivers then received messages to help reduce stress for 1 month. After the month was over, the Annie coordinator completed a program evaluation with the caregivers to obtain their feedback on the messages.






Ethics Approval

The pilot evaluation was reviewed and approved by the Memphis VA Medical Center institutional review board (IRBNet #1415769). It was conducted from July 11 to November 1, 2019, to evaluate caregivers’ use and satisfaction with the messages and format.

Stress Management Caregiver Protocol Development

The Annie Stress Management protocol for caregivers, used in the pilot and the national rollout, is based on the Resources for Enhancing All Caregivers Health (REACH) caregiver intervention used in the Department of VA and the community [4,29,30]. REACH and the text messages are based on the stress health process model [31,32]. As conceptualized by the model, caregivers have challenges and demands placed on them (eg, care of their loved one, lack of help, and their own physical health) and cognitive and emotional responses to these challenges (eg, grief, feeling alone, and viewing events from a negative perspective) [32]. If caregivers do not believe they have resources and the capacity to manage their demands, they experience stress, which can lead to physical and psychological distress and illness [31]. Following the model, the Annie Stress Management text messages focus on knowledge, strategies, and actions that caregivers can take to cope with demands and their responses to them and manage stressors. The goal is to help the caregiver intervene at multiple points in the stress health process (Table 1).

Table 1. Stress health process model and Annie message examples.

Model	Annie message examples
Challenges and demands placed on caregivers 	<ul style="list-style-type: none"> “Getting enough sleep & rest will help you cope and feel better. Tell your doctor you are a caregiver and ask for help.” “Annie here. When you need help or a break from providing care for your loved one it’s ok to let friends or family members know.”
Emotional and cognitive response to challenges 	<ul style="list-style-type: none"> “View stressful situations from a more positive perspective. See a traffic jam as a chance to listen to music or enjoy some alone time.” “Expressing what you’re going through can help even if you can’t change a stressful situation. Talk to trusted friends or go see a therapist.”
Resources and the ability to manage challenges 	<ul style="list-style-type: none"> “Keep a stress journal. Make note of when you experience stress to see if there is a pattern. Find ways to remove or lessen those triggers.” “Feel like no one gets you? Try a support group. Ask your Caregiver Support Coordinator https://www.caregiver.va.gov/support/New_CSC_Page.asp”
Perceived stress 	<ul style="list-style-type: none"> “Anxiety can be managed. Healthy eating, less caffeine & alcohol, relaxing, & time with friends can help. Maybe try meditation or yoga, too.” “Take a deep breath. Gently reach your arms to the side, then reach them out in front of you. Now reach up as high as you can. Repeat.”
Emotional and physical response Illness 	<ul style="list-style-type: none"> “As a caregiver you’re at risk for high blood pressure, heart problems, colds, & flu. Be sure to watch your own health.” “Some caregivers don’t have energy for routine tasks. If this sounds like you, seek tips from your doctor or other caregivers.”

Caregiver Stress Management Pilot Intervention

The pilot protocol included two types of text messages, those that Annie sent without any caregiver involvement focusing on motivation and education and messages that were requested by the caregiver after a series of prompts by the system and caregiver responses. For the pilot messaging workflow, motivational or educational messages were sent twice a week without any caregiver involvement. The requested messages offered an optional activity to the caregiver. Caregivers were contacted to ask if they were stressed twice a week (on days when a motivational message did not come). If they replied yes, they received a query to ask if they would like to do an activity to help. Caregivers could respond to this activity-requested query to receive content about stress-relieving strategies, breathing, give yourself a break, setting boundaries, and mindfulness. Annie then sent messages to help with stress. For example, for breathing, one stress-relieving strategy was “Annie says to breathe deeply, hold it for 3 seconds. Breathe out slowly. Say a calming word to yourself. Let your jaw, shoulders, and arms go limp. Repeat twice.” Caregivers could contact the text program at any time as many times as they wanted for stress-relieving messages. Each time they texted the appropriate prompt to the system (eg, Activity BREATHING), a message was sent until the bank of 10 to 12 texts for each topic was exhausted and began again. Therefore, caregivers could receive none, one, or several activity messages. Annie provided instructions each time to help the caregivers remember how to request assistance.

Evaluation

At the end of a month, caregivers were contacted by telephone to evaluate acceptability of the program’s texts by the coordinator. Questions were focused on technical aspects (eg, easy to read and problems requesting texts) and perceptions of benefit (eg, helped me take better care of myself, made me feel cared for, and made me feel confused). Each question was answered on a scale from 1 (completely disagree) to 5 (completely agree). Data were analyzed using descriptive statistics, including frequencies and means. Open-ended questions asked caregivers what they enjoyed about the messages and why, usefulness of the messages, most helpful messages, use of the messages, and confidence about managing stress after the messages. Caregivers were also asked whether they liked the request prompt messages (responding yes to being stressed and receiving an activity message).

Quotes were examined individually by 2 anthropologist authors, both with prior experience in coding of qualitative data. Each reviewer sorted the descriptions, concepts, and central ideas into potential themes [33] looking for repetitions, similarities, and differences [34]. Topics that occurred repeatedly were linked to verbatim quotes [33]. Themes were discussed and finalized by these authors.

Results (Pilot)

Of the 29 eligible caregivers, 22 (76%) caregivers of veterans from 9 facilities returned consent forms and were enrolled in the study. Only one caregiver identified as a male. Baseline demographics for the 22 caregivers showed an average age of 57 years. Most caregivers (18/22, 82%) were White individuals.

All the 22 caregivers reported multiple diagnoses for their veterans; the most common reported diagnoses were posttraumatic stress disorder (PTSD; 18), dementia (7), and depression (7). These figures are similar to caregivers of veterans nationally, where 96% are female and 61% are at least aged 50 years, and the top two diagnoses reported for the veterans are mental illness and PTSD [35].

In addition, 82% (18/22) of the caregivers provided follow-up data (Table 2).

Qualitative data yielded 3 themes. Caregivers reported that the messages and activities were helpful and helped them manage stress:

Used breathing exercises—those were great. Take time for myself to calm down and breathe, then rethink the situation.

One caregiver said the following:

I don’t always know what to do when I’m stressed. Reading the messages calms me down. Focuses it elsewhere. Liked the directions to destress. Don’t always think about that.

Caregivers also reported that the texts made them feel like someone cared for them and that they were not alone, which is evident from the following testimony from one caregiver:

Don’t have much family. Nice to know someone cares I’m alive.

Finally, the texts made caregivers take care of themselves and think about their own needs:

I felt that for once it mattered about me. I mattered. I could stop and really think about myself instead of just going through the motions of everyday life.

Comments indicated that caregivers tried the strategies. One caregiver reported about a mindfulness and PTSD distraction activity with an ice cube in the hand:

My favorite activity was the ice one. It made me stop and just watch. Had the Vet do it too when he was upset. Got him to calm down.

The caregivers also felt that the messages were easy to read and convenient, which can be gleaned from the following comment:

I shared with my friend. They were easy to access. Could go back to them as needed.

The caregivers’ responses did not indicate that the messages caused them difficulty.

Three caregivers felt that they had received too many messages. When caregivers were asked about responding to the messages about being stressed and then requesting an activity, 86% (12/14) of the caregivers who answered preferred not to request an activity:

Send out something positive instead of asking for it and ask how your stress level is.

Another caregiver reported that they did not like to think about stress:

Love just getting the messages. Don't like thinking about stress. Asking if I was stressed brought it to the forefront. Don't always realize I'm stressed.

Another caregiver commented the following:

Just give an activity. If you're in a stressful state, you don't know what to do. You're stressed about everything. You don't know what you want/need.

Caregivers were also busy:

Like just getting the messages is easier for me with taking care of my husband and baby.

Another said the following:

Just send an activity. I get busy and don't always realize I'm stressed.

Table 2. Annie Stress Management protocol pilot evaluation responses (n=18).

Responses	Completely disagree, n (%)	Disagree, n (%)	Neutral, n (%)	Agree, n (%)	Completely agree, n (%)	Mean (SD)
1. The texts helped me manage my stress.	1 (6)	1 (6)	3 (17)	9 (50)	4 (22)	3.8 (1.1)
2. I feel this program helped me take better care of myself.	2 (11)	1 (6)	4 (22)	5 (28)	6 (33)	3.7 (1.3)
3. I felt like someone cared about my personal well-being when I got the texts.	1 (6)	2 (11)	2 (11)	3 (17)	10 (56)	4.1 (1.3)
4. I would recommend this service to another caregiver.	2 (11)	1 (6)	0	2 (11)	13 (72)	4.0 (1.7)
5. It was easy to read the texts.	0	0	1 (6)	2 (11)	14 (78)	4.5 (1.2)
6. I received the texts at awkward times.	8 (44)	3 (17)	3 (17)	3 (17)	1 (6)	2.2 (1.4)
7. Receiving the texts interfered with my daily life.	15 (83)	3 (17)	0	0	0	1.2 (0.4)
8. I was confused when I received the texts.	17 (94)	1 (6)	0	0	0	1.1 (0.2)
9. I had problems sending in keywords and/or making responses.	7 (39)	8 (44)	1 (6)	1 (6)	1 (6)	1.9 (1.1)

Text Messaging Caregiving Intervention (National Rollout)

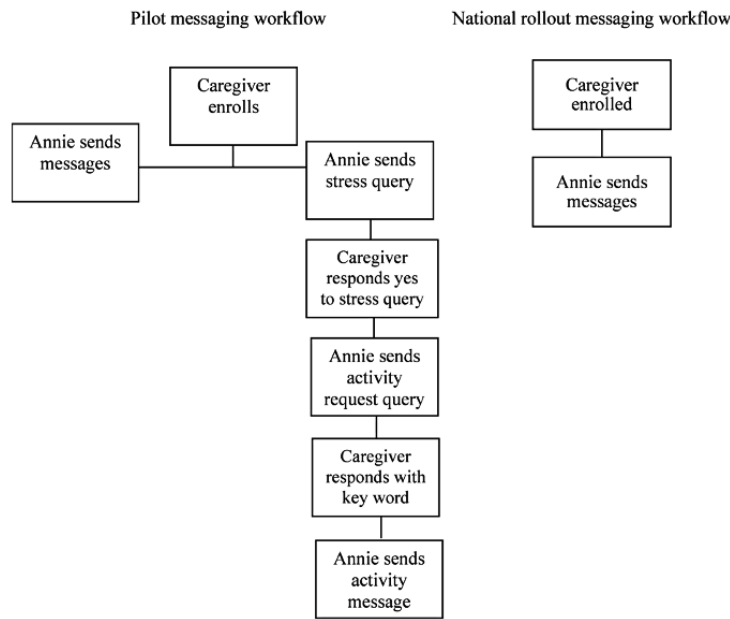
Methods (National Rollout)

Overview

On the basis of the positive response to the messages by caregivers in the pilot study, a national rollout of the Annie caregiver stress management protocol was approved by the VA's Office of Connected Care for the VA system on October 1, 2019. As the caregivers from the pilot study had expressed a strong preference for being given an activity instead of requesting it, modifications to the protocol were made to have Annie offer messages without the necessity of caregivers replying as shown in [Figure 1](#).

The program was initially publicized through the Caregiver Support Program and the Office of Connected Care of the VA. The eligible candidates were caregivers of veterans enrolled in the VA health care system who had a VA electronic health record or a veteran who was a caregiver. Any VA staff member could register a caregiver from their facility in Annie. Staff at VA facilities determined whether a caregiver was interested in receiving the Annie text messages. If the caregiver was interested, staff then ensured that caregivers had a VA electronic health record, which was a requirement for Annie services, registered the caregiver in Annie, and sent a referral to the Caregiver Center for the caregiver to be entered into the Annie protocol. The Caregiver Center offered training to staff who did not know how to use Annie, and as of February 2022, a total of 592 staff members had been trained.

Figure 1. Pilot and national rollout SMS text messaging workflow.



Annie Stress Management Protocol

The Annie caregiver stress management protocol was lengthened to include a year’s worth of messages. On the basis of pilot findings, 3 messages per week, education, motivation or inspiration, and activity were sent with no response required from the caregiver.

In the current version of the protocol, messages are sent 3 times a week on different days and focus on education, motivation or inspiration, and strategies or skills (Table 3). Educational texts provide information about stress and responses to it. Motivational texts, often quotes from famous people, were

included on caregiver request and were validating and inspiring. Activities focusing on stress-relieving strategies, breathing, giving yourself a break, taking care of yourself, setting boundaries, and mindfulness are strategies suggested for caregivers to try. General messages were sent to all caregivers in addition to the protocol messages. Messages offering an email link for help or questions and links to the national Caregiver Support Program staff and resources were sent every 3 weeks. Caregivers were registered in Annie by clinical staff at their local VA facility; each caregiver must have an electronic health record. After registration, a referral was sent to the Caregiver Center, which enrolled the caregiver in the requested protocol.

Table 3. Annie Stress Management protocol message examples.

Educational (Tuesday 10 AM)	Motivational (Thursday 3 PM)	Activity (Saturday 10 AM)
“Stress is normal in life- it’s how we react to threats. Let us help you develop effective ways to manage stress.” [Annie]	“In order to carry a positive action, we must develop a positive vision.” [Dalai Lama]	“Inhale for 4 seconds; hold for 6 seconds; exhale for 4 seconds; hold for 6. Repeat 5 times.” [Annie]
“Holidays can be overwhelming. Avoid taking on too much. Keep things simple. Ask for help. You may need to reduce activities.”	“Annie here. It’s important to remember that you are doing a good job taking care of your loved one. Keep it up!”	“Go outside and enjoy the sun and the breeze for a few minutes.” [Annie]
“Annie invites you to listen to a monthly talk about caregiving. https://www.caregiver.va.gov/support-line/presentations.asp ”	“The only limit to your impact is your imagination and commitment.” [Anthony Robbins]	“Each day & week do something with your loved one that gives you both pleasure.” [Annie]
“Annie here, with a reminder: Making your family the priority is nothing to feel guilty about.”	“Thank you for taking care of your loved one. Even if no one remembers to tell you, what you are doing is appreciated and makes a difference.”	“Write down 3 things you’re grateful for.” [Annie]

Evaluation

At the end of 6 months, caregivers were asked to fill out a brief evaluation survey through a SurveyMonkey (Momentive Inc) link in an Annie message. All data were anonymous. Six questions asked caregivers’ opinions about the texting program using a 5-point Likert scale ranging from completely disagree (score=1) to completely agree (score=5), and data were analyzed with descriptive statistics, including frequencies and means.

Caregivers were asked whether the texts had helped manage stress, increase confidence, and take better care of self and loved one. They were also asked whether the texts had made them feel like someone cared about their well-being and whether they would recommend the program.

Two open-ended questions about additional topics and anything caregivers wanted to add were asked. Quotes were examined individually by 2 anthropologist authors, both with prior

experience in coding of qualitative data. Each reviewer sorted the descriptions, concepts, and central ideas into potential themes [33] looking for repetitions, similarities, and differences [34]. Topics that occurred repeatedly were linked to verbatim quotes [33]. Themes were discussed and finalized by these authors.

Results (National Rollout)

Overview

In total, 4401 caregivers were enrolled in the stress protocol as of the end of February 2022. All had not been in the protocol for 6 months and were therefore not eligible to receive the evaluation link, with 3522 caregivers eligible. Figure 2 shows the responses from the 781 caregivers who answered the link, a 22.17% (781/3522) response.

Caregivers endorsed 3 messages per week as the right amount (679/778, 87.2%). The mean scores for each question indicated

that the caregivers felt that the messages had been useful in increasing their confidence (mean 4.1, SD 0.9), managing their stress (mean 4.3, SD 0.8), and helping them take care of themselves (mean 4.3, SD 0.8) and their loved ones (mean 4.2, SD 0.8). The messages also helped them feel cared for (mean 4.5, SD 0.8). Finally, caregivers would recommend the program to other caregivers (mean 4.5, SD 0.8). In fact, several caregivers reported sharing Annie even on social media. Furthermore, 23.1% (180/778) of the caregivers tried all the strategies, an additional 42% (329/778) tried most strategies, and 26.9% (210/778) tried some strategies. Less than 10% (49/778, 6.3%) tried a few strategies and only 1.3% (10/778) of the caregivers tried none of the strategies.

In their open-ended comments, the caregivers identified 5 general themes, as shown in Table 4. There were only 16 negative comments; most wanted to respond and have Annie answer them (ie, wanted a real person checking on them), and 2 caregivers thought there were too many texts.

Figure 2. Annie national rollout caregiver stress management text protocol evaluation responses.

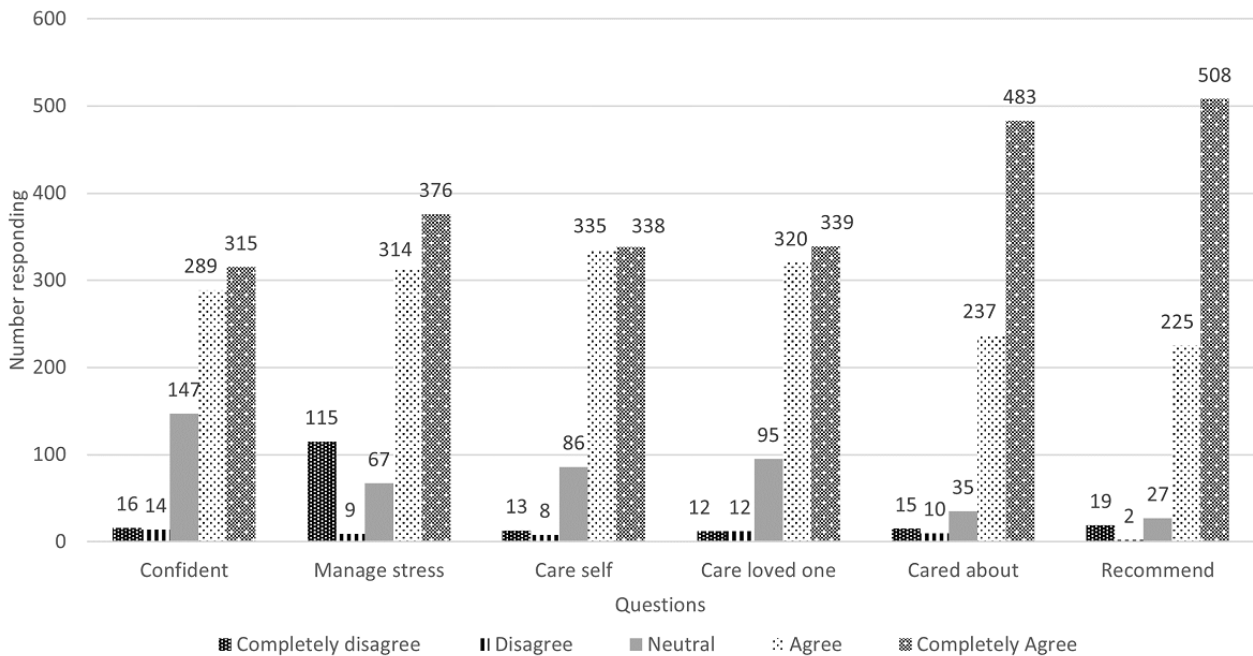


Table 4. Annie benefit themes and quotes.

Themes	Illustrative quotes
Appreciate texts; bright spot for me; were helpful; shared with others	<ul style="list-style-type: none"> • “I so look forward to Annie. It is a bright spot in my day and helps tremendously.” • “The texts have been very helpful, and I know I have needed them more than I knew. And I share many of the messages with my fellow caregiver. I so appreciate having a lifeline.” • “These tips and quotes are perfect and truly help my sanity and remind me to breathe.” • “I share Annie with my sister who is also a caregiver for her husband and daughter. Annie always seems to address whatever we happen to need help with. We both love receiving texts from our ‘special friend’.”
Right on target; came when I needed them	<ul style="list-style-type: none"> • “Sometimes it was right on target for my emotions and when I least expected it.” • “The messages that were given were inspiring and uplifting seemed to come always at the right time. Loved the messages that were personal and uplifting as simple as look at you, you’re doing awesome. Those really helped me out at times and seemed to come at just the right moments. So thank you so much for those!!”
Help with self-care	<ul style="list-style-type: none"> • “The messages were/are a welcomed reminder to ‘self-care.’ Thank you!!!!” • “These texts for me were great reminders to take better care of myself and not be too hard on myself. Thank you!” • “Annie reminds me when I forget myself sometimes. Thanks Annie!”
Keep them coming; want them to start again	<ul style="list-style-type: none"> • “Just continue sending them...thank you” • “VA Annie is just what I need to make my day. Keep them coming!” • “Just keep giving me the reminders that I will be able to get us both through this.”
Show me I’m not alone; someone cares	<ul style="list-style-type: none"> • “Just that I am not alone.” • “I just like to know that someone cares.” • “The messages always come at the times when I need to know I am not alone.”

Poststudy Experience

Because caregivers shared the protocol with their veterans, and staff were asking whether veterans could be put on the protocol, a year-long Annie Stress Management protocol for veterans was developed, which is similar in format and types of messages. In February 2022, this protocol enrolled 294 veterans, making it one of the top 10 protocols according to the number of users.

On the basis of the success of the stress management protocol, several new protocols have been developed: bereavement; taking care of you, which focuses on healthy lifestyle and emotions; dementia behaviors, which combines stress management and management of behaviors; and PTSD behaviors, which focuses on coping with behaviors. Similar to the stress management protocol, education and strategies for all these protocols are also taken from the REACH VA behavioral intervention. All protocols except bereavement last for a year. Caregivers can also be enrolled in the VA’s Coronavirus Precautions protocol.

The Annie caregiver protocols had a cumulative total of 7274 caregiver enrollments as of February 2022 with 7062 current users, and each month about 250 caregivers are referred to the Caregiver Center for enrollment in an Annie protocol, showing the feasibility of incorporating SMS text messaging into a large health care system. The Annie caregiver protocols have been extremely successful with more users than many Annie Veteran protocols. For example, in February 2022, among the top 10 Annie protocols based on users, 3 were caregiver protocols. Of the 6386 users in the top 10 protocols, 56.34% (3598) were enrolled in caregiver protocols with 31.99% (2043) of the users enrolled in stress management.

Discussion

Principal Findings

In this evaluation of the Annie Stress Management texting pilot and national rollout for caregivers of veterans, caregivers felt that the messages had been useful in increasing their confidence, managing their stress, and helping them take care of themselves and their loved ones. The messages also helped them feel cared for and made them feel less alone, looking on Annie as a friend. The caregivers felt that the messages were right on target and did not want them to stop. These findings are similar to those that veterans have articulated about Annie [27,28].

During the national rollout, caregivers’ requests led to some changes in the stress management protocol, namely, the last week of all the caregiver protocols let the caregiver know that the protocol was ending, thanked the caregiver, and provided the link to request an extension or a new protocol.

Part of the success of the Annie caregiver protocols may be because of their cross-diagnosis applicability and the fact that neither caregivers nor clinicians must respond to the messages. The impact on staff workload does influence the uptake [36]. Research into veteran and staff use of Annie suggests that additional support helps providers adopt, implement, and sustain the program [36,37]. The Caregiver Center has provided this support through coordinator to promote the protocols, help providers with the technology, and train them to use the technology. In addition, because caregivers often rely on others to identify useful technology [6], the relationship between caregiver and VA staff likely facilitates caregiver’s willingness to try SMS text messaging.

One surprising finding was that caregivers in the stress management pilot study reported that they did not want to ask for an activity. In reviews of web-based caregiving interventions, interaction with peers or professionals or interactive support has been shown to be beneficial [7-10]. Despite the lack of 2-way interaction with a clinician or with Annie in the final national rollout protocol, caregivers still found benefit and felt cared for. There may be several reasons for this finding. Caregivers may be too busy for protocols that ask for a response, or they may not want to respond when they are not responding to and receiving answers from a real person. As the text message comes directly to the phone, the messages may feel personal and intimate as a text message from a friend would. The Annie caregiver text programs have positive motivational messages and jokes, two features that are unlike most Annie Veteran facing protocols, which may add to a feeling of being personal. Caregivers did tend to anthropomorphize Annie, thanking her for being there for them.

Our study has some limitations. The lack of negative findings may reflect that people who did not like Annie might have asked for it to be turned off before receiving the 6-month survey link to provide feedback. Another limitation is that the SurveyMonkey data are anonymous; therefore, caregiver characteristics cannot be linked to satisfaction or outcomes. Finally, the 22% response rate is low.

Conclusions

Although many caregiving interventions are multicomponent and targeted to specific issues of the caregiving dyad [1-4], the stress health process model shows that education, support, and strategies and resources to manage challenges and cope with stress [31,32] are important components of successful interventions. As has been shown with the Flo and Annie texting protocols, these are all part of what users can receive through

texting, leading to increased confidence building and empowerment [24,27,28]. For caregivers specifically, research has shown that having concerns acknowledged, perceived attention, and positive regard can all be therapeutic [38], suggesting that any positive contact around caregiving can be beneficial.

This type of SMS text messaging provides a means of reaching many caregivers and is practical for health care systems or clinical practices. Cloud-based messaging systems could be developed with relatively low hosting and per recipient costs. Moreover, texts provide a way to overcome the lack of widespread implementation of interventions into clinical settings, which is a critical barrier to improving outcomes for caregivers and the loved ones they support [1,3,39]. For caregivers, there are similar areas of concern, including self-care, emotional and physical well-being, communication, and stress management. Health care organizations could provide the service to multiple types of caregivers either with or without condition-specific messages, which would be more closely targeted to the dyad's needs. Text messages can be an adjunct to more traditional therapeutic techniques [40] or used on their own without the caregiver needing to respond to a clinician or to the system. These text messages are perceived as a caring touch from the organization:

To know I'm not alone and that you're thinking of us. Caregiving is very lonely, so a phone call or text saying, hey, how are you today? I'm thinking about you makes a big difference.

Caregiving messaging, incorporated into clinical settings, represents a seamless, low-cost way to provide useful and meaningful support to caregivers, who frequently feel overlooked by the health care system.

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Authors' Contributions

JLMA and LON conceptualized the intervention and drafted the article and accepted direct responsibility; CDC and JRM made substantial contributions to the intervention and collected, analyzed, and interpreted data and contributed important intellectual content; and CRH made substantial contributions to the intervention and important intellectual content.

Conflicts of Interest

None declared.

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Abbreviations

PTSD: posttraumatic stress disorder

REACH: Resources for Enhancing All Caregivers Health

VA: Veterans Affairs

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Original Paper

Decision-making for Parents of Children With Medical Complexities: Activity Theory Analysis

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Abstract

Background: Shared decision-making (SDM), a collaborative approach to reach decisional agreement, has been advocated as an ideal model of decision-making in the medical encounter. Frameworks for SDM have been developed largely from the clinical context of a competent adult patient facing a single medical problem, presented with multiple treatment options informed by a solid base of evidence. It is difficult to apply this model to the pediatric setting and children with medical complexity (CMC), specifically since parents of CMC often face a myriad of interconnected decisions with minimal evidence available on the multiple complex and co-existing chronic conditions. Thus, solutions that are developed based on the traditional model of SDM may not improve SDM practices for CMCs and may be a factor contributing to the low rate of SDM practiced with CMCs.

Objective: The goal of our study was to address the gaps in the current approach to SDM for CMC by better understanding the decision-making activity among parents of CMCs and exploring what comprises their decision-making activity.

Methods: We interviewed 12 participants using semistructured interviews based on activity theory. Participants identified as either a parent of a CMC or a CMC over the age of 18 years. Qualitative framework analysis and an activity theory framework were employed to understand the complexity of the decision-making process in context.

Results: Parents of CMCs in our study made decisions based on a mental model of their child's illness, informed by the activities of problem-solving, seeking understanding, obtaining tests and treatment, and caregiving. These findings suggest that the basis for parental choice and values, which are used in the decision-making activity, was developed by including activities that build concrete understanding and capture evidence to support their decisions.

Conclusions: Our interviews with parents of CMCs suggest that we can address both the aims of each individual activity and the related outcomes (both intended and unintended) by viewing the decision-making activity as a combination of caregiving, problem-solving, and seeking activities. Clinicians could consider using this lens to focus decision-making discussions on integrating the child's unique situation, the insights parents gain through their decision-making activity, and their clinical knowledge to enhance the understanding between parents and health care providers, beyond the narrow concept of parental values.

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KEYWORDS

shared decision-making; activity theory; parental decision-making; parenting; participatory medicine; pediatric; caregiving

Introduction

Children with medical complexity (CMC) are defined as individuals with complex chronic disease necessitating specialized care, high family-identified needs, functional disability, and high health care utilization [1,2]. As of 2007, it was estimated that CMC in Ontario, Canada comprised 10% of all hospital admissions and approximately one-quarter of hospital days [3]. The population of CMC is a heterogeneous one, including diverse medical conditions such as brain injuries, cerebral palsy, or extreme prematurity, conditions that are severe and complex due to the intersection of multiple organ systems being affected [4]. Due to their complex care needs, the demands on parents and families of CMC are high, with parents of CMC interacting, on average, with 13 different physicians and specialists representing 6 subspecialties [3]. In the United States, it is estimated that caregivers of CMC spend 11 hours to 20 hours per week coordinating the care their child receives from their multiple providers [5]. As a result, parents of CMC become intimately familiar with both the health care system and their child's specific health care needs, as active participants in the provision of care.

Given the challenges of medical care for CMC, their parents are faced with many difficult decisions. Unlike parents of healthy children who seek occasional care for a broken bone or an acute respiratory illness, parents of CMCs often face a continuous number of interconnected decisions, often without the support of medical evidence due to the complexity and co-existence of multiple chronic conditions [6,7]. To support these complex decisions, shared decision-making (SDM), a collaborative approach to reach decisional agreement, is a proposed means of improving health outcomes for children with chronic medical conditions [8,9]. However, the application of SDM in pediatrics and for CMC specifically is still poorly understood [10] and underpracticed when compared with children without medical complexity [9].

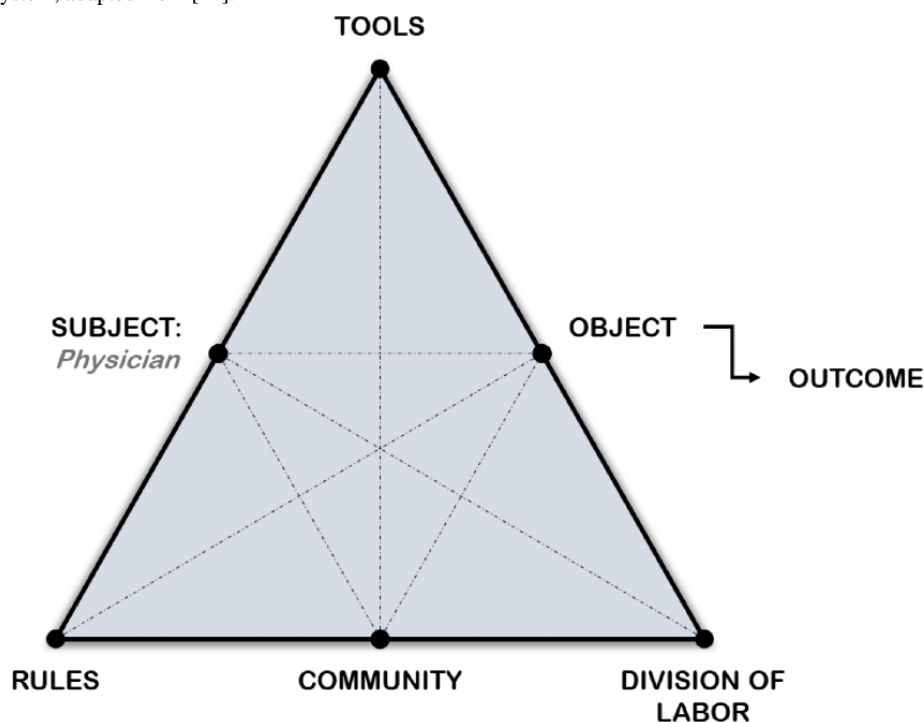
Frameworks for SDM have been developed largely from the clinical context of a competent adult patient facing a single

medical problem, presented with multiple treatment options informed by a solid base of evidence [7,10]. However, given that this is not the case for CMC, parents may undertake the decision-making process differently than adults facing a discrete medical choice. The goal of this study was to explore the decision-making of parents of CMC as an activity within the context of a process shared between clinician and parent but external of current SDM frameworks.

Methods

Overview

In this qualitative study, activity theory informed both the data collection and analytical approaches taken. The semistructured interview method used in this study is based on the critical decision method (CDM) 5-step plan [11,12], supplemented with probes focused on the elements that comprise an activity as laid out in activity theory [13,14] using the Activity-Oriented Design Method (AODM) [15]. The interviews were analyzed using activity theory as a guiding framework and applying framework analysis methods [16-18]. A cross-disciplinary framework for studying different forms of human practices, activity theory provides a framework to view individual and social systems as interlinked, continuously evolving processes [19]. Activity theory was selected as the framework to guide both data collection and analysis as it provides a map that outlines the elements that comprise a human practice or activity considering an individual's action, reactions, reasoning, and behavior with a broader context of influential rules, beliefs, and practicalities. The elements of the activity theory system as pictured in Figure 1 consist of (1) those involved in achieving the aim (Subjects), (2) the mediating artifacts used in the activity (Tools), (3) the rules that govern the activity (Rules), (4) other actors involved in the activity (Community), and (5) the division of activities among actors in the system (Division of Labor) [20]. The study was approved by the Research Ethics Board of the University of Toronto, and signed informed consent was obtained prior to each interview.

Figure 1. The activity system, adapted from [21].

Sample and Participant Recruitment

We recruited 12 participants (10 mothers, 1 father, and 1 young adult, formerly [child] with medical complexity) via social media groups for parents of children with medical needs in Ontario, Canada. The inclusion of a young adult CMC was a pragmatic decision, as the young adult joined the interview with their parent to elaborate on the story and provide additional insights. Eligibility criteria included being English-speaking, 18 years or older, and caring for a CMC. To determine if the child qualified as a CMC, each prospective parent participant completed a questionnaire listing the criteria for medical complexity ([Multimedia Appendix 1](#)). Parents who answered “yes” to at least 2 criteria for medical complexity were included in the study. Qualifying diagnosis for the CMC included children diagnosed with neurological disorders including cerebral palsy; rare diseases; and complex respiratory issues, including those requiring a tracheostomy and mechanical ventilation. Recruitment ended when thematic saturation was achieved. Saturation was reached when no new elements of the activity system, tools, rules, community, subject, object, and division of labor ([Figure 1](#)) were identified. The sample size is similar to studies utilizing similar methodologies [22,23].

Data Collection

Data were collected using semistructured interviews based on CDM [11,12]. CDM is a type of cognitive task analysis interview and knowledge elicitation technique that consists of a 5-step semistructured interview plan with specific knowledge elicitation probes [12]. CDM’s 5 steps consist of (1) select incident, (2) obtain unstructured incident account, (3) construct incident timeline, (4) decision point identification, and (5) decision point probing. Interviews lasted 1 hour to 2 hours and were conducted in person by a single interviewer (FB). Participants were asked in advance to prepare a story about a

time they had to make a difficult decision regarding the medical care of their child in consultation with their child’s medical team. The parent started the interview by relaying the story without interruption. To further elicit detailed information on “the judgements, assessments, and decisions” [11] along with the “motives, social and cultural issues within the context of the activity” [15], the interviewer used follow-up probes adapted from CDM [11] and the activity theory–informed AODM [15]. Notes were taken during the interview outlining the timeline of the decision, and keywords linked to each probe were documented. Interviews were also recorded and transcribed verbatim by a professional transcriber.

Analysis

Interview transcripts and notes were analyzed using the 5 steps of the framework analysis approach developed by Ritchie and Spencer [16–18], namely familiarization, thematic analysis, indexing, charting, and mapping and interpretation. During the familiarization and thematic analysis phases, 2 researchers (FB, CL) independently read and open coded the same 3 transcripts looking for emergent themes and activity theory concepts (tools, community, rules, division of labor, object, and subject) [13]. A final coding scheme was developed by jointly discussing disagreements and reaching consensus on the themes and activity theory concepts identified in the data. At the indexing phase, the final coding scheme was used by the first author (FB) to re-code all 10 interviews in Nvivo 12 (QSR International; Burlington, MA). During the charting phase, relationships were established between codes, and similar codes were grouped together. Using Mwanza’s 8-step model [15] as a guide, the themes and activity theory elements were mapped to the activity triangles ([Figure 1](#)), and different activities were identified based on their objective (object within the activity system). Focusing on each activity and related subactivity as the unit of analysis, interpreting the data consisted of annotating the relationships

between the elements that comprise the activity, noting the tensions, contradictions, and actions embarked upon to overcome them. The results of the analysis were sent to all interviewees to comment on the analysis and interpretation of the data as to further corroborate the findings. Feedback from interviewees included confirmation that the findings reflected their lived experience and suggestions to revised quotations that they believed would reveal their identity. Suggested changes were incorporated and approved by interviewees.

Results

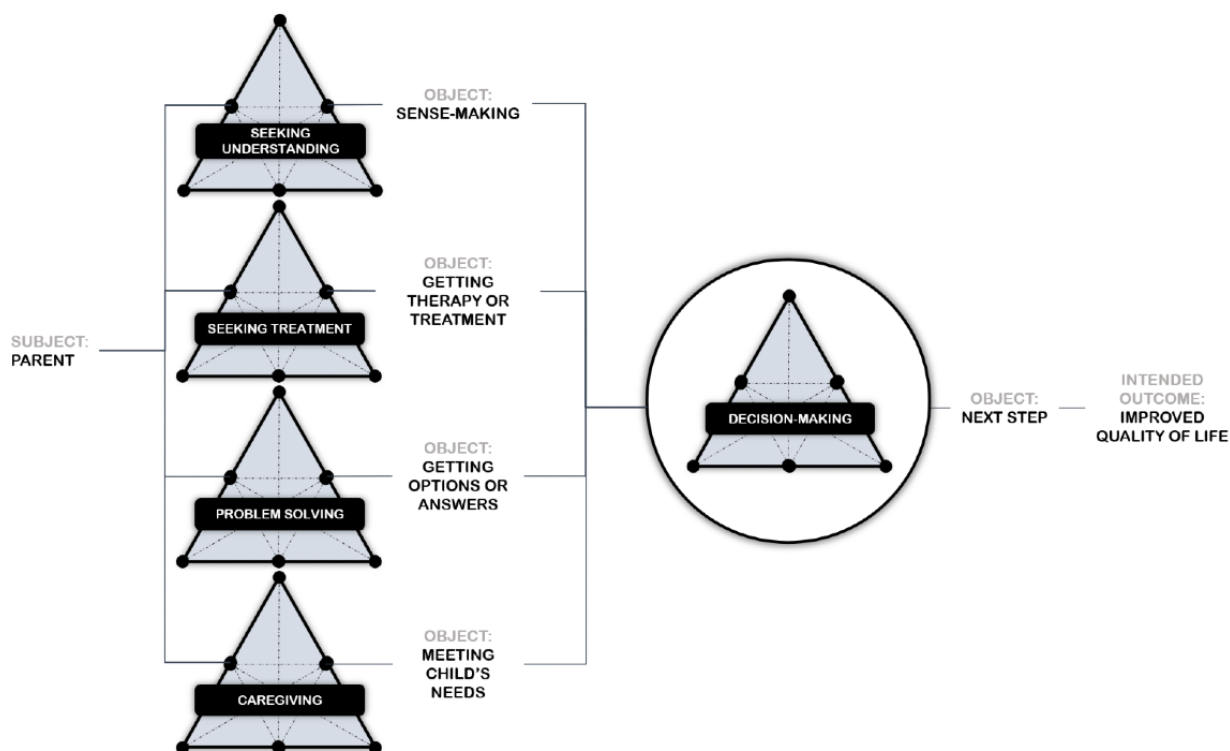
Primary Interview Findings

The interviews conducted relayed stories about difficult decisions that ranged from the appropriateness of a surgical intervention to decisions around admission to hospital. Although the difficult decisions being discussed varied in terms of interventions, they were similar in that all were deliberated over multiple conversations with input from multiple health care providers. The decisions were also similar in that they all had a long-term goal of improving the child’s quality of life.

Our analysis of the parental decision-making process identified that the activity was comprised of 4 subactivities, outcomes of which were inputs into the larger decision-making process. Figure 2 depicts the relationship between the 4 activities of (1) seeking understanding, (2) seeking treatment, (3) problem-solving, and 4) caregiving and the larger decision-making activity. As noted in Figure 2, each subactivity was oriented toward different distinct immediate goals (object) that were necessary to achieve the outcome of the larger decision-making activity. For parents of CMC, decision-making was not just a single cognitive process of weighing the risks or choosing between available options. Rather, by engaging in the 4 subactivities, parents make sense of the context in which a decision is being made while also experimenting with problem-solving solutions to develop rules that govern and inform future decisions.

The narrative of our results in the following sections consists of describing how each subactivity unfolds, including an explanation of the actions taken by the parents (subjects) to overcome challenges that occur within the process.

Figure 2. The parent’s decision-making activity system.



Subactivity 1: Seeking Understanding

The activity of seeking understanding was present in all the interviews and characterized by the need to seek out information to support the parents’ understanding of the situation they were facing. The aim (object) of this activity we label as “sense-making” as the activity is directed toward interpreting the situation as to transform it [24].

Because we really didn’t know. Like there is not a lot of information we were given. We kinda had to do our own research, figure it out and what not. [3002]

The activity of seeking understanding is initiated by the parents being presented with a problem or decision point for which an answer was unclear.

We were presented with the idea after about two days after her heart surgery that we should try and extubate her and you know, not having I guess a full understanding of what that would mean for her or what that would look like because of the lack of knowledge around her lung and heart function. [3004]

Parents engaged in the activity of seeking understanding when they found themselves unclear on how their child’s specific

context could affect the outcome. For this activity, the subjects (parents) used a series of mediating tools, such as journal articles, test results, and Facebook to make sense of their child's condition and formulate questions that, when answered, would improve their understanding.

These tools were validated against other cognitive and behavioral tools such as past experiences and inquiring questions. Access to health care providers in team meetings (community) also facilitated getting answers. The activity of seeking understanding involved questioning if the information being provided applied to their child's specific needs.

The strategy going forward from there was to increase his medication, but they were doing it sooo slowly...I got around to actually looking at the literature myself on pediatric dosing, I was frustrated again because it was so low...the nurse who was doing the prescribing kept trying to, "He is on a lot of doses...I think he metabolizes it very quickly." And the nurse had us try and go to the "normal" number of doses. Why? Why did she do that? [3006]

Depending on the situation, the activity was focused on understanding why a health condition was occurring, why a specific suggestion was made, or the evidence to support a proposed course of action.

I want to understand things, I want to be spoke to in layman's terms, I don't like a lot of medical jargon that confuses me...I want them to dumb it down for me so I feel comfortable and I feel informed and I leave with the security of knowing that she's going to...that I know. She may not get better, but I just need to be in the know. [3012]

Activities of understanding were both successful and unsuccessful. Barriers that limited the subject's ability to understand included gaps in the availability of information (tools), medical jargon, or systemic barriers to accessing the right people to answer the parents' questions (rules). At times, these challenges led to an unfulfilled activity, resulting in uncertainty.

I don't really know a lot about what the options are because with our last conversation, we didn't really get a lot of information because we stopped the meeting because they realized that the key players were not in the room. [3011]

A common barrier to understanding was not being involved in the discussions with doctors.

They [the doctors] weren't involving us in any of their decisions. They were making decisions that we didn't know that they were making without understanding the risks and benefits involved and without informing us of any risks that they understand that we didn't understand...They would talk about whatever they would talk about, and they would come back with their decision, and we just weren't involved. [3006]

Not being involved in discussions left a gap in parents' understanding of the reasoning and deliberations that led to a conclusion:

Just because someone tells you they have expertise does not mean that they're using it properly and does not mean they have expertise in your child. [3006]

The final decision or recommendation, even from doctors with extensive credentials, was not sufficient to support the parent's ability to make sense of the situation.

When the activity was successful, the outcome was knowledge that informed further activities. When the activity was met with challenges or remained unsuccessful, those challenges were overcome by undertaking a secondary activity such as problem-solving or repeating the activity of seeking.

By making sense of their circumstance, parents felt more comfortable making a decision they felt was the correct one.

I didn't have to think twice about it. You know, because everybody was already there, everybody gave their input. Here it's like we get a little more information from this person but if they're talking without the other person being there, so it's like, would you say the same thing around the other person, right? So, I remember that was, when we made the decision, I made the decision by myself, I didn't even tell my husband. [3002]

In contrast, when families felt that they were blocked from gaining a full understanding of their CMC situation, they were unsure if the options and opinions put before them were the right ones. Parents wanted to understand why an option was put forward by clinicians, including the factors the clinicians considered and whether all available options were included in the deliberation.

I don't know if we had exhausted all the measures to get the information that we needed around her heart. [3004]

Gaps in the information or lack of appropriate tools to obtain a full understanding impeded the desire to conclude the decision-making activity.

Subactivity 2: Seeking Treatment

The activity of seeking treatment is one where the parent either actively embarked on seeking out an intervention or passively agreed to the intervention suggested by the doctor and undertook the tasks to acquire it. The act of seeking out treatment took a large portion of each parent's time, and much of that time was devoted to obtaining the treatment.

It wasn't a difficult decision for us, it was difficult to get it to happen, it was difficult to get the doctors to decide to have it happen. [3006]

Parents were driven to seek treatment or tests as an activity to obtain a solution to the identified problem. The outcomes of the activity were sought to provide input into a larger decision or as a tool to aid in the decision-making activity.

Parents developed their own set of tools to move the activity forward and overcome barriers. Persistence was a common tool utilized by parents in repeatedly engaging with health care providers. They often adapted their communication styles or

the way they presented the situation based on how well the technique (or tool) has worked for them in the past:

I kept calling the secretary's office, put us on a [surgery] cancellation list, put us on a cancellation list... Well I've come to learn sometimes you need to, uhmm, this is in air quotes "exaggerate the situation." We said, well this isn't really exaggerating but to the secretary it might have sounded [starts whispering] worse than it was. [3001]

Taking on the role of advocates for their children, or as parents often framed it "I would push, I would push again and push harder" (3004), was what parents deemed necessary to overcome the barriers to accessing services, even though many parents did not want to take on such a role.

I didn't like the position I was in, in that I had to tell the doctors to do their job. But I didn't mind it, I had no qualms with telling them... [3012]

However, when services were offered, parents felt more comfortable taking on more of a passive role within the division of labor.

They just sort of said, here's the, this is the surgery, this is the surgery that he needs. And we at first said, okay, I guess if that's the surgery he needs, that's the surgery he needs. [3011]

A common theme stated by parents was that they felt the need to trust their physician's ability to balance evidence with the specific needs of the child. Parents were comfortable with taking a passive role only if they trusted that the options put forward were based on the specific needs and considerations of their child, after doctors have researched the full suite of options available.

Even so, our analysis did not identify that parents of CMC were aware of any tools that were used by doctors to convey their deliberation process. The result was a tension between the parents' desire to trust the physician to execute their job (Division of Labor) and their need to validate that the physicians' actions were based on the child's specific needs and not other conflicting motives or influencing factors, including standard hospital rules or protocols.

So, I'm frustrated with the clinicians, why wouldn't they tell you what are the options, do they not know these exist? I highly disbelieve that the neurologist who works at "Hospital A" doesn't know about a gait lab, that her colleague runs. Why didn't she tell me about this? ... Why won't she say, hey, how about you go see a movement specialist? Do they not know, do they not want to tell us, are they overloaded and bombarded themselves that, you know, we're just another number for them, they just want to move on to the next appointment? [3001]

Access to available treatment options was an identified barrier dictated by the rules, community, and division of labor within the activity system. For example, rules requiring doctors' referrals for certain procedures at times limited obtaining or changing therapy and treatment.

They fully said they will not do this procedure. At one point, I finally got to say, there is nothing I can do to change your mind on this? [3005]

When the activity of seeking treatment was met with barriers, the outcome for parents was often frustration or uncertainty. For example, parents of CMC were frustrated that physicians controlled access to interventions because of the lack of reasoning provided.

They just said "no that is not how we do things." That's an exact quote. I will never ever forget it. [3006]

The lack of information required to support decision-making drove parents to seek out other alternatives, such as embarking on a problem-solving activity of their own, repeating the seeking activity, or looking to understand the situation with insufficient tools (information). The barriers and facilitators identified in this activity informed how parents embarked on the decision-making activity or related activities. Parents looked to treatments available from accessible sources, such as accommodating physicians or peers:

Everything I asked for, she [the doctor] accommodated. Whereas sometimes if you ask a doctor for a certain test, they just disregard it and say they don't need it. She was very open to ordering everything I asked for. [3010]

When barriers arose, tools such as persistence were sometimes not enough, and luck often played an important role in gaining access to care.

I ran into our [specialist]...and she asked me how it was going, and I burst into tears (laughs) and then she helped us out. I don't mean I like [made] a rational phone call and requested help. It was desperate times. [3006]

The outcomes of this activity were not only the results of therapy but also knowledge of how the system's rules work, development of the parents' beliefs of their role, and knowledge on best sources of treatment options, which may not always be the physician.

This tacit knowledge of the system or observations from the therapy were integrated into the decision-making activity as best practices (tools), for example, always booking appointments with the same clinician to ensure consistency and continuity of care.

When we go to the clinic, we always schedule with the same orthoptist, we always schedule with the same ophthalmologist. That the vision clinic at "Hospital A" has like four or five different ophthalmologists, we specifically request the same clinicians so that, because their notes are consistent, they see the trend, they know, you know what I mean, instead of flip-flopping within the clinic. [3001]

Subactivity 3: Problem-solving

The activity of problem-solving is a process of trial and error, experimentation, and hypothesizing. Parents referred to the activity as their role or responsibility, which was required due

to gaps in service provisions or for collecting evidence to support decision-making. The activity of problem-solving is oriented toward finding an answer to, or reason for, a specific problem with an aim toward achieving a longer-term goal (outcome) such as understanding options to present to clinicians.

The problem-solving activity is comprised of connecting and using tools such as test results, journal articles, social media, past personal experience, and observations.

We had worked really hard to learn as much as we could about the condition, to talk to our faith leaders, to talk to other parents, to talk to anybody and everybody we felt would be wise and to get as much of a sense of, like, we knew that we wouldn't be able to answer all of the questions beforehand, but we wanted, like, a working theory on how we were going to answer the questions. [3005]

Parents also reached out to community members such as peers, doctors, and family for assistance and insights.

Facebook, social media is how I learn everything. [3001]

Social media provided parents with the opportunity to connect with peers who may also have experienced similar problems.

The activity was framed and influenced by rules such as the availability and access to tools or community members (clinicians or peers) willing to share their own experiences, which could be used as tools. Some parents referred to their own educational backgrounds as nurses, health care administrators, doctors, engineers, or basic scientists, which influenced how they viewed the problem but also provided them with skills to access and evaluate tools, such as journal articles. Having access to specific skills drove how they proceeded with the problem-solving activity but was also seen as something not visible or valued by health care providers.

I wish [health professionals] wouldn't assume that all parents get their information from Google and Facebook, because, yes, obviously, I joined all the possible Facebook groups for parents of children with cerebral palsy and so on. But I also know how to use PubMed, I looked at, and I got my husband who is a doctor, and I got my husband to look at things with me. I feel like I've done the academic research, but I've also done the parent perspective side, because when you go on Facebook groups, people talk about these things like SDR surgery and what sorts of questions should I be asking, and what was your experience? I feel like I've covered both the real lived experience, and I also try to cover the academic evidence-based side. But health professionals always assume that parents just go on Facebook, or they say things like, well, stay off Google. Well, Google is not a bad starting point. It's not somewhere you should necessarily end, but it's not a bad starting point. I think health professionals actually need to give parents more credit because, yes, parents read everything, and a whole bunch of what they read

might be irrelevant, but they also might read some stuff that's valuable or relevant. [3007]

In the problem-solving activity, the parents' intended goal was sometimes fulfilled and at other times resulted in frustration when barriers were encountered. In one interview, the family explained how a limitation of access to medical equipment limited their ability to trial their solution, causing them to be frustrated as they attempted to get help from a clinician.

So, we kept thinking about it and trying to deal with it and we came up with this hypothesis that he was [health condition] and then we went to [the specialist] to ask if, we could maybe try [intervention] to see if [the intervention] would stop the [condition/symptoms]. [3006]

The influence of the rules or barriers resulted in outcomes that were at times different from the intended ones. This resulted in either frustration, uncertainty, or in gaining knowledge or experience. The resulting outcome drove the next activity such as seeking, caregiving, or making a decision with the newfound information or hypothesized solution.

Subactivity 4: Caregiving

The activity of caregiving was represented in all 10 interviews. Participants described how they managed doctors' appointments, delivered medical care, observed medical problems, and tried to keep their child happy and healthy. In the activity of caregiving, parents learned about their children, responded to their needs, and documented their progress. Like a detailed medical chart, some parents collected and collated years of data as part of the caregiver activity:

We had all the results there, we had all of the names of all the doctors there, we, I could give them [child's name] birth weight, I could give them their weight at a year, I could give them their weight at two years, I could tell them every infection they had, like they couldn't have asked for any more detail than we had. [3005]

These parents did not embark consciously on a data collection activity. Rather, in the act of caregiving, they identified barriers to accessing data already collected in medical encounters and imbedded collection or collation of data as part of that activity. Similarly, gaps in training or knowledge were identified during the caregiving activity.

I realized that I could tell when he was having an apnea episode very easily and I could rub his back and that would get him breathing again. But like nobody explained to me any of these things. [3005]

The act of caregiving was mediated by a variety of tools, such as access to information on how to provide proper care, personal observations, and knowledge gained from past experiences. The activity of caregiving is a continuous cycle of using tools to help decide what to do, observing the outcome, then re-examining the child's condition to inform next steps. If the outcome was negative, the activity was reoriented to one of the related activities to obtain new knowledge and tools to continue the process.

Rules were a major driver of the activity of caregiving. The rules imposed by the child's medical condition and treatment, such as specifically timed medications or use of a ventilator to maintain life support, all drove the act of caregiving, sometimes causing stress:

There was no leisure, there was no going and doing anything. And then the stress that we were under ...all the time was just crazy. [3006]

These rules, motivated by medical needs, drove the need for caregiving tasks and restricted the ability to do the task but also drove the desire to find solutions to ease the burden of these tasks.

We had him vented 24/7 again...[but] we knew that it didn't have to be this way...So we weren't really like invested in figuring out how to move around with the vent. We were invested in getting him off the vent. [3006]

In the act of caregiving, parents identified changes in their children's needs but also identified gaps in how health care providers addressed those changes. Hospital rules that silo care and limit interdisciplinary and team-based care drove parents to take on the role of care coordination to overcome this barrier. Coordination was a role frequently cited in the interviews, as parents were able to view the full picture of the child's care, whereas health care providers only saw pieces of it:

I'm the one who takes care of all her care. I'm the one who knows all of the moving pieces. I'm the one who is with her every day. Other parts of her team see her maybe once every few months. They don't know the day-to-day of what she's going through and what impact things will have on her. [3011]

This kind of episodic care, born out of how hospitals are structured (rule), drove parents to act as coordinators of care. When the activity unfolded well, parents were happy that their child was living a fulfilled life. When the activity was met with barriers and outcomes were not achieved, it could cause frustration and uncertainty. Irrespective of how the activity unfolded, a secondary outcome of the caregiving activity was gaining experience and confidence but also a feeling that their expertise was not valued enough by health care professionals.

I know X [child name] at her best, I know X at her worst. I know X in-between. I'm able to gauge if the concern should be high. I am able to gauge if the concern should be low. I know X, I know every single thing about that little human being, that they just don't know. They know [disease], but I know X's [disease], and every kid's [disease] is different. They sometimes, they're just too stuck in their textbook definitions of what [disease] is, but X's version of [disease] is what I know, and so that makes me an expert that they don't give enough credit to. [3012]

Outcomes of the caregiving activity were identifying decisions to be made, validating parents' roles in the decision-making activity, and providing the knowledge to support the decision. The knowledge of a child's reactions to medication, therapy, or treatments was an outcome of the caregiving activity, which

became an input to the decision-making activity. From coordinating care, parents also collected, cross-validated, and documented information from multiple clinicians. This information then became a tool in the decision-making activity. In addition to tools, the caregiving activity provided parents with a sense of their role in the decision-making process and with confidence to make an informed decision or identify their knowledge gaps.

Discussion

Principal Findings

Frameworks of SDM in pediatrics are evolving. What was once viewed as the process of supporting a patient and their caregiver in choosing between multiple treatment options has now incorporated the understanding that the complex reality of making decisions is underserved "by depicting the making of 'a' decision as a discrete act" [7]. The findings of our study support recent findings from Feudtner et al [7] that decision-making by parents of CMCs consists of multiple decisions that shape and inform future care decisions. By using an activity theory lens, our study identified that parents of CMCs make decisions based on a mental model of their child's illness, informed by the activities of problem-solving, seeking understanding, obtaining tests and treatment, and caregiving. Our findings depict the parental decision-making process as a continuous process connecting the parent's past, with decisions made in the present and future.

Whereas previous studies have identified a multitude of influences affecting parental decision-making, including "cultural norms, community standards, impact on siblings or extended family, previous experiences, religious faith, and impact of acuity and stability of the child's health status" [8,25], our study instead focused on how these background elements, combined with systemic rules and beliefs in the participants' roles, drive actions and decisions. The activity theory framework and the probes developed from previous work completed by Hoffman et al [11] and Mwanza [15] focused the interview on identifying the needs and the activities undertaken to fulfil them. The framework of activity theory deconstructed the complexity of the decision-making task into smaller pieces (elements within the activity system) [13] that could be analyzed to determine the relationships between elements and how they evolve over time. Using activity theory as the structure for the data analysis not only organized the tasks that comprise the larger activity but also revealed the distinct short-term goals (objects) that informed the actions that parents embarked on, thereby informing the larger activity (eg, caregiving).

Proposed frameworks for SDM in pediatrics are still grounded in the belief that the goal of SDM is to improve medical outcomes for children by combining parental values with current evidence [8-10]. However, the findings of our study suggest that the basis for parental choice and values brought to the decision-making activity are developed via activities looking to build concrete understanding and capture evidence to support their decisions. What has been conceptualized as parental values in pediatric decision-making models are in fact tools developed from parents' activities, which serve to support the larger

decision-making activity. Parental beliefs and values described in SDM models are identified in our study as concrete tools that include findings from experimentation, behaviors learned from prior medical encounters, and observations gained from performing caregiving tasks.

These tools, which others have presented as heuristics, “ease the tasks of decision-making because they fit unfamiliar, complex, or novel information into familiar patterns of thought and language. By using common maxims and rules of thumb, parents can tackle the current challenges of decision-making by casting the daunting situation in terms and concepts that in the past have helped to make sense of other situations, solve problems, and communicate” [26]. Our study has taken steps to identify the activities that develop the heuristic tools identified by Renjilan and colleagues [26] and show that the activities that create them are an integral part of the decision-making activity.

The activities parents complete to formulate their decisions are important to understand in-depth when developing solutions to improve SDM for CMC. For example, in the act of problem-solving, parents formulate a hypothesis or potential solution to the problem that they bring with them to decision-making deliberations. However, without tools for doctors to explore these hypotheses and how they were formulated, these are often excluded from decision-making discussions and minimized to parents’ values. Additionally, our findings that parents embark on an activity to understand their child’s medical reality are key to addressing gaps in current

SDM models. Acknowledging that parents make decisions based on an understanding that is constructed by engaging in concrete actions, rather than just developing abstract values, further supports the importance of parental contribution to the decision-making process as active participants. The findings of our study detail the specific activities performed by parents that build their sense of empowerment, expertise, and knowledge. An important next step in this area of work is further empowering parents with the knowledge that the activities they perform are important and valued in the SDM process. Practice recommendations outlined in Table 1 provide examples of how clinicians can support empowered SDM by incorporating the findings from this study into SDM conversations.

Our study has several limitations. First, this research focused on the decision-making practices of parents only and did not consider the perspectives of the physicians involved in the decision-making. Although the rich narratives we obtained provide insight into parent’s actions and reasoning for those actions, our study relied on retrospective accounts using a single data collection method. To mitigate the potential for recall bias, future research may apply additional methods, such as observations of parent-physician encounters in situ. Second, we chose to focus on CMC as it is a population with extraordinary health needs who are supported by caregivers that are generally highly invested in the health of their children. Thus, the perspectives of parents of CMC in our study may not be generalizable to parents of other pediatric populations.

Table 1. Practice recommendations for clinicians embarking on shared decision-making (SDM) for children with medical complexity (CMC).

Key findings	Practice recommendations
Parents make decisions based on their lived experience: Parents of CMC use information collected from the daily acts of care such as problem-solving, seeking understanding, obtaining tests and treatment, and caregiving to inform their decision-making. As active participants in the delivery of care, parents of CMC develop their expertise as caregivers and gain a valuable knowledge to inform decision-making.	Empower parents by acknowledging that the daily activities they perform in caring for their child are the basis for their expertise as caregivers and a valuable source of knowledge to inform decision-making. When seeking parental perspectives to inform SDM, direct questions toward parental knowledge, actions, and observations parents have made, rather than only their long-term goals or broad values they may hold.
Understanding is contextual: When trying to understand their child’s medical condition (sense making) parents endeavor to gain a sense of how their child’s specific context could affect the outcome. Parents want to trust that the options presented by the physician are based on the specific needs and considerations of their child.	When presenting medical options for care, provide background and reasoning in relation to the child’s specific needs, family context, the larger body of options considered, and known evidence base. Consider connecting parents with peer families to facilitate discussions that may address practical, social, and community issues grounded in lived experience.
Multiple activities influence decision-making: Parents make decisions based on the completion of multiple activities including caregiving, problem-solving, obtaining treatment, and sense making.	Be mindful of the needs of parents that may fall outside of immediate decision deliberation but still impact how decisions are made (eg, vacation time for parents considering a surgical intervention). Provide a supportive environment to discuss all aspects of care related to the decision-making process including the outcomes of, caregiving, problem-solving, obtaining treatment, and sense making. Consider tools and resources that can support the decision-making process outside of clinical encounters.
Rules guide and influence activity outcomes: Rules such as cost of therapy or medication can limit the number of options available to parents. Parents make decisions fully aware of these limitations.	Be aware of rules or structures that may be limiting the ability of parents to fulfill the options presented to them and address them openly (eg, presenting options that are too expensive).

Conclusion

When viewing the decision-making activity as a combination of the caregiving, problem-solving, and seeking activities, we can address both the aims of each individual activity and the related outcomes (both intended and unintended). Understanding that the outcome of problem-solving is a carefully crafted idea

or hypothesis should focus clinicians on questioning what occurred in the problem-solving activity to develop that idea. When addressing how to educate parents on the medical options, it could be useful to view the parents’ seeking understanding as a sense-making activity aimed at bridging the gap between their current situation, specific to their child’s personal context, and desired outcomes [27,28]. This view could help clinicians

focus conversations toward integrating the child's unique situation with knowledge gained from general standards of care and help reach greater understanding between parents and health care providers, beyond the narrow concept of patient (or parental) values. Challenging the belief that, in SDM deliberations, patients and families bring values and physicians bring clinical expertise, similar to other studies [29], our findings

show that parents are active participants in the delivery of their child's health care. Thus, viewing the information and insights gained from the caregiving, problem-solving, and seeking activities as broader than values should inform physicians to engage with the information provided by parents as a form of expertise.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Eligibility questionnaire for a parent of a child with medical complexity.

[[DOCX File , 18 KB - jopm_v14i1e31699_app1.docx](#)]

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Abbreviations

- AODM:** Activity-Oriented Design Method
CDM: critical decision method
CMC: children with medical complexity
SDM: shared decision-making

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Original Paper

In Anticipation of Sharing Pediatric Inpatient Notes: Focus Group Study With Stakeholders

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Abstract

Background: Patient portals are a health information technology that allows patients and their proxies, such as caregivers and family members, to access designated portions of their electronic health record using mobile devices and web browsers. The Open Notes initiative in the United States, which became federal law in April 2021, has redrawn and expanded the boundaries of medical records. Only a few studies have focused on sharing notes with parents or caregivers of pediatric patients.

Objective: This study aimed to investigate the anticipated impact of increasing the flow of electronic health record information, specifically physicians' daily inpatient progress notes, via a patient portal to parents during their child's acute hospital stay—an understudied population and an understudied setting.

Methods: A total of 5 in-person focus groups were conducted with 34 stakeholders most likely impacted by sharing of physicians' inpatient notes with parents of hospitalized children: hospital administrators, hospitalist physicians, interns and resident physicians, nurses, and the parents themselves.

Results: Distinct themes identified as benefits of pediatric inpatient Open Notes for parents emerged from all the 5 focus groups. These themes were communication, recapitulation and reinforcement, education, stress reduction, quality control, and improving family-provider relationships. Challenges identified included burden on provider, medical jargon, communication, sensitive content, and decreasing trust.

Conclusions: Providing patients and, in the case of pediatrics, caregivers with access to medical records via patient portals increases the flow of information and, in turn, their ability to participate in the discourse of their care. Parents in this study demonstrated not only that they act as monitors and guardians of their children's health but also that they are observers of the clinical processes taking place in the hospital and at their child's bedside. This includes the clinical documentation process, from the creation of notes to the reading and sharing of the notes. Parents acknowledge not only the importance of notes in the clinicians' workflow but also their collaboration with providers as part of the health care team.

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KEYWORDS

medical informatics; information sharing; electronic health records; patient portals; hospitals; pediatrics; focus group; inpatient care; caregivers

Introduction

Background

In 2001, the Institute of Medicine acknowledged that the “free flow of information” between patients and electronic health

record (EHR) systems is central to the principle of patient-centered care [1]. Tang and Lansky [2] proposed that patients need access to their personal health information, at minimum “their own diagnoses, medications, allergies, lab test results, visit summaries, and other findings over time.” They

further argued that access to this information could enable patients to enter into a true dialogue with their health care team, increasing not only their understanding of the treatment required but their motivation to engage in such treatment—essentially establishing themselves as the locus of control in the relationship.

Patient portals are a health information technology that allows patients and their proxies, such as caregivers and family members, to access designated portions of their EHR using mobile devices and web browsers [3]. Patients are now potential readers and users of EHRs. This has redrawn and expanded the boundaries of medical records. In this way, patient portal technology has been deemed a “digital disruption” in the health care industry—a “type of environmental turbulence induced by digital innovation that leads to the erosion of boundaries and approaches that previously served as foundations for organizing the production and capture of value” [4]. Sullivan and Staib [5] reported that over 50% of EHR implementations fail because organizations do not appreciate the degree to which such digital transformations can be disruptive. These authors further identify various “syndromes” associated with digital disruptions, including digital deceleration (reduced efficiency) and hypervigilance (anxiety and overreaction in the face of change) [5].

To facilitate the success of digital transformations in health care and mitigate disruption, a mutual understanding of health information exchange and relevant technologies is required by all stakeholders involved—patients and their caregivers and health care professionals. The objective of this study was to investigate the anticipated impact of increasing the flow of EHR information, specifically physicians’ daily inpatient progress notes, via a patient portal to parents during their child’s acute hospital stay—an understudied population and an understudied setting.

The Open Notes and Copying Letters Initiatives

Patient access to personal health information in the United States was greatly accelerated in 2010 by a patient-centered movement called Open Notes. The collaborative experiment that launched the movement involved Beth Israel Deaconess in Massachusetts, the Geisinger Clinic in Pennsylvania, and the Harborview Medical Center in Washington State. At these 3 centers, 20,000 patients were invited to read their ambulatory visit notes written by their clinicians using their patient portals. Results were overwhelmingly positive, with 59% to 77% of patient survey respondents agreeing that viewing their clinicians’ notes helped them feel “more in control of their care” [6]. Since then, 51% of US adults who accessed their medical records via web in 2020 reported that these records included clinical notes [7].

The Copying Letters initiative, which began in the United Kingdom in the 2000s, presents an interesting and relevant initiative parallel to Open Notes. Launched in April 2004, this initiative was similarly grounded on the idea that all patients should carry a summary of their medical record [8]. To enable this summary to be as complete as possible, all clinicians were required by the National Health Service to send their patients copies of all letters they had written about them, for example, a letter describing their case in the context of a referral to

another specialist [9]. The cited benefits of this practice were very similar to those articulated in the Open Notes movement. Supporters argued that, by providing access to the contents of the record, Copying Letters put the patient “at the centre of care” [10] and effects “a shift in the balance of power” [11].

Advocates of medical record transparency argue that there are many additional benefits, including enhanced physician-patient communication, improvements in patient understanding of their own condition and ability to perform self-care, and increased patient engagement and participation [12,13]. Therefore, this transparency has now been mandated by law in the United States under the 21st Century Cures Act. Effective April 2021, a total of 8 types of clinical notes—consultation, discharge summaries, history and physicals, imaging, laboratory reports, pathology reports, procedure, and progress notes—must be shared with patients [14].

Pediatric Inpatient Context

Although the Open Notes and Copying Letters initiatives have both been adopted at a national level, only a few studies have focused on sharing notes with parents or caregivers of pediatric patients. Early studies of Copying Letters were conducted because some pediatricians were concerned about the effect of sharing clinical content on adolescent patients and parent readers [15]. They feared not only that these readers would be confused but that any sensitive information might be stigmatizing or offensive. These studies were built on the early work by Partridge [15] who explored parental reactions after reading their child’s pediatrician’s assessment reports.

The body of Copying Letters research repeatedly documents the ways in which parents, as in-home managers of their children’s health, perceive themselves as silent partners of physicians [15-18]. It is clear that these parents valued access to clinician-authored documentation of their child’s care. A very early study by Partridge [15] found general satisfaction with pediatricians’ letters among parents of children living with disabilities, with 74% of parents being satisfied with what they read. Other researchers found similarly high rates of satisfaction among patient readers. Cowper and Lenton [17] reported: “One hundred percent of parents were pleased to have received the letter” from their child’s pediatrician. Liapi et al [19] compared adult patients in an otolaryngology clinic with 100 parents of pediatric patients; 77% of the parents found the copied letters helpful. Most recently, Amirav et al [18], who surveyed parents of pediatric patients with asthma, reported that 80% of the parents called the letters “helpful” and 98% indicated that they would want similar letters in the future.

In the United States, the Open Notes movement began with adult outpatients. Researchers have only now started to investigate the access behaviors of inpatients and their reactions to content, ranging from medication information [20] to their full medical record [21]. A review of the medical literature reveals a small but growing body of literature on access to medical records by hospitalized patients but finds even less research in pediatrics [22]. This mirrors the general situation for EHRs and personal health records, in which the research literature largely concerns adult outpatients [23,24]. Therefore, there are significant gaps in our understanding of patient- and

caregiver-facing tools in the context of inpatient care [25], particularly Open Notes in the pediatric hospital setting. Instead, much of the research published on young patients focuses on the complexities of policies surrounding access to patient portals by children and adolescents [26]. One research group at the Boston Children's Hospital designed a framework for a system of personally controlled health records to be accessed by "parents, guardians, and third-party entities" while remaining in the patient's control [27].

Kelly et al [28,29] were the first to investigate the use of an inpatient portal by parents of pediatric patients, increasing our understanding of parental motivations for accessing their child's inpatient records in real time. Of the 14 parents interviewed in that study, 13 were interested in having access to physicians' notes in the portal:

I don't know that doctors necessarily keep it a secret, but in my son's entire medical history, I've only had one doctor really turn the screen to me and sit there and say like "Here's what we're seeing, here's what's happening." So, if I could see things like [notes] in here, that would be amazing. [Parent]

Parents suggested that notes would provide a recapitulation of information, serve as a memory aid, and improve their understanding and ability to advocate for their child:

Sometimes talking is different than writing. Sometimes I will forget the point. [With notes], we'll know where's the problem and what's the next step. [Parent]

When you read, you can understand it much better. [Parent]

Others suggested that they would like to refer to notes when they were unavailable during morning rounds:

I wasn't here [during rounds]. So, if they say the doctor's notes are on there, I could be able to read them and see what [the doctor's] suggesting. [Parent]

However, some parents had concerns that notes could cause undue anxiety and had reservations about the impact of sharing on physicians:

I don't know how comfortable [doctors] would feel. It may feel like an invasion of [doctors'] privacy. [Parent]

This study builds on these early findings and continues our systematic approach [30] to evaluate the perspectives of key people potentially involved in this digital disruption—the sharing of physicians' notes with families of hospitalized children. These findings will allow for a mutual understanding of stakeholder perspectives and facilitate the success of note sharing in light of recent federal mandates.

Methods

Study Design and Participants

This qualitative study was conducted at a Midwest academic children's hospital between October and November 2018. A total of 5 in-person focus groups were conducted with 5 different types of stakeholders considered most likely to be impacted by

sharing of physicians' inpatient notes with parents of hospitalized children. There were no exclusion criteria; 4 groups were composed of hospital staff with the roles described later, whereas parents were recruited from the hospital's Patient and Family Advisory Council, a standing committee of volunteers. Separate focus groups were conducted for each stakeholder role to encourage participants to respond freely, without the fear of retribution. Adolescents were intentionally excluded from the focus groups because of the complexities of access to adolescent health information.

Focus group participants were recruited via email. All participants were provided with an information sheet describing the study and risks and benefits. Informed consent was obtained; participants were not reimbursed.

Ethics Approval

This study was approved by the institutional review board of the University of Wisconsin-Madison (protocol ID number: 2018-0913).

Focus Groups

Each group met in a private conference room for 1 session; the sessions lasted 1.5 to 2 hours and were audio recorded. Using a semistructured facilitator guide consisting of open-ended questions, moderators asked all focus group members for their opinions about the potential of providing parents of patients aged ≤ 12 years with real-time access to daily inpatient progress notes using a bedside tablet during their child's hospitalization. To facilitate the discussion, moderators provided an example of a daily progress note and reviewed the general content of these notes with all focus group participants. Participants were then asked to reflect on sharing progress notes with parents.

Data Analysis

Audio recordings of focus group sessions were transcribed by a professional service, and transcripts were deidentified and coded using Dedoose (version 8.3.17, SocioCultural Research Consultants, LLC). Three researchers participated in coding using a constant comparative method [31,32]. Two researchers (MMK and CAS) independently reviewed all transcripts and met with the third researcher to develop a codebook. These 2 researchers then coded all transcripts and consulted with the third researcher to reach consensus concerning any discrepancies, always referring back to the transcripts [33]. The themes were summarized and presented using illustrative quotes. Further details about the study methods are available in a study by Smith et al [30].

Results

Demographics

The 5 focus groups comprised 6 *administrators* (leaders in the hospital and residency program, information services, risk management, and patient relations), 7 *pediatric attending hospitalist physicians* (physicians whose primary professional focus is on hospitalized patients), 5 *pediatric intern and resident physicians*, 8 *bedside nurses*, and 8 *parents* who had experience caring for a child in the hospital. A total of 34 participants were included in the 5 groups. These participants were largely White

(30/34, 88%) and female (27/34, 79%) and held college degrees (23/34, 68%).

Benefits of Inpatient Open Notes

Distinct themes identified as benefits of pediatric inpatient Open Notes for parents emerged from all the 5 focus groups. These themes were communication, recapitulation and reinforcement, education, stress reduction, quality control, and improving family-provider relationships.

Communication

The value of Open Notes for improving communication between the inpatient health care team and members of the patient's family was commented on by various focus group participants, but particularly by residents. They also saw notes as a way to improve communication between parent caregivers:

The one potential benefit that I can think of is that in these families that have, say, four children, and one of them is in the hospital, so both parents can't always be there,...Dad or Mom, if they have to stay home that day, can read the note from that day... [Resident]

Communication between members of the health care team and the patient's family could potentially be enhanced. Multiple participants saw value in making the treatment plan accessible before rounds to increase families' understanding and potentially change the family dynamic or discussion with the team:

[M]ost of the questions you get overnight are related to the plan...What are we doing?...What's going to happen tomorrow? [W]hat are we waiting for? [T]he plan is something that may benefit [them], and their having it may reduce the questions. [Resident]

One parent who had been able to view her child's physician notes during a hospital stay at another institution pointed out that seeing the notes gave her information about communication, which was another benefit:

I've had the good fortune to see some of the doctor's notes...it allows you to learn a little bit more about what's going on...Did so-and-so understand, or did I understand what was being said? [Parent]

Recapitulation and Reinforcement

Nurses pointed out the usefulness of Open Notes as a tool to empower families with information, relieving parents of the need to ask hospital staff clarifying questions:

[F]amilies know too that we, as nurses, are busy and physicians are busy...later in the day, they could be talking to Dad or another family member, and it just gives them a tool...to be able to speak to and look back without having to necessarily bother us. Because, a lot of times, that's what they say. "Oh, well, we didn't want to bother you."...[I]t would give them an extra tool to look back... [Nurse]

Parents valued the idea of Open Notes for providing families with a text-based source of information that reiterated and reinforced what had already been relayed verbally. This was important for recapturing knowledge in the short term:

[P]art of the objective would be to talk about goals and getting released from the hospital, things like that. Sometimes those are multistep, and there's a lot there, and it's hard to remember just from a verbal conversation. [Parent]

It was also valuable to access this information over time:

[H]aving that at your fingertips is, it's so much easier when you have to...remember down the line something for the school, or something for a social worker...that you could quick go back and look at...where was he on this scale when he did his neuro test? [Parent]

Education

A hospitalist commented on the potential value of notes for families as an educational intervention, deployable for people in different learning situations:

[F]amilies should probably end up having better understanding, better health literacy as a result of this, because they will have the words that they missed when someone was talking too fast or...in an accent, or using words they've not heard before, that they can now look up at their leisure without feeling embarrassed about asking questions that they weren't sure they should ask. [Hospitalist]

Stress Reduction

A mother described how a visible plan would provide her with structure to reduce her anxiety:

For me, my biggest issue with my mental state and my anxiety around my daughter is when something is going on and there's no plan. I feel like I'm trying to reach someone, you know...trying to get in, trying to be seen, and like there's that question mark. I don't know if it's serious. I don't know if it's not serious...I...automatically feel more at ease as a parent when I know that there's steps that we're going through to improve the situation. Like there's a roadmap. [Parent]

A nurse voiced her opinion that showing families the breadth and depth of information being collected about the patient would itself serve to lower parental stress:

I think we could eliminate some of the anxiety of the parents just reading that...explaining, we don't think it's this, but we are going to rule out this, this, this to make sure that we're covering all of our bases. [Nurse]

Quality Improvement

Unsurprisingly, hospital administrators talked most about Open Notes' potential for improving the quality of health care delivery:

...I think what [Open Notes] will also do is prompt further discussion...if there's information in the medical record...that a family doesn't understand or that we've written incorrectly, that's in the medical record now. And so...it's almost like another set of

eyes on what we're thinking about the path for either the patient or, in this case, our parents.
[Administrator]

However, both hospitalists and parents also commented on the potential for Open Notes to be used as a mechanism for quality control, describing the potential value of parents in improving the accuracy of medical records:

Hopefully, [parents will] feel like they know the plan better. Maybe they're going to check, they're going to see that something is inaccurate. [Hospitalist]

[T]here might be critical pieces of information that may not have been stressed enough or could be missed in that period of rounds, and so it gives you the opportunity to say, hey, this other topic...that was really important to me. And only with a second set of eyes would you be able to capture that information...I think it's really important. [Parent]

Improving Family-Provider Relationships

Hospital administrators argued that Open Notes could play a role in reassuring parents, and one parent agreed:

Sometimes you're only in the room, so maybe five, ten minutes, but it's actually a very complicated case...I am in the room a short time, but my note is extensive. That could give [parents] more reassurance that I did, in fact, think about all the stuff that maybe we didn't talk about...But they're like, wow, that person really is thinking about my case.
[Administrator]

Once I have steps in place, we're going to check this, rule this out, move onto this, I...automatically feel more at ease as a parent when I know that there's steps that we're going through to improve the situation. [Parent]

Challenges of Inpatient Open Notes

The focus group participants also pointed to the particular challenges posed by inpatient Open Notes. The 5 dominant themes were burden on provider, medical jargon, communication, sensitive content, and decreasing trust.

Burden on Provider

The most frequent challenge of Open Notes was the idea that transparency and access to notes by parents would place an undue burden on hospital providers. Every provider focus group mentioned this theme, particularly the residents:

[R]ight now, we work 16 hours...and now if you're going to add on top of that having to run to the parents' bedside to explain our note, that's going to delay all our other responsibilities. [I]f you are going to add extra documentation...that's going to be more work that we aren't necessarily going to have time to do. [Resident]

One hospitalist acknowledged adding the patient's family as a new reader of the note and spoke about the extra work involved in considering an additional audience during the writing process. They anticipated that learning to write notes for parental viewing

could be challenging for residents who were still honing their note-writing skills. They used the analogy of a parent teaching a child how to write:

[W]hen you have your child who's writing something, you ask them to go back and edit themselves...that is an expectation. And if they're having trouble with that, then you say this is the checklist of things you need to look for. Does every sentence have a capital? Does every sentence have a piece of punctuation on it?...Are all the words spelled correctly? So, in a similar vein, we almost need a checklist for the residents to say,...have you done this, have you done this, is this accurate, before you submit it to me. Because that would also potentially reduce the amount of time I'm going to spend on doing it.
[Hospitalist]

In addition, one member of the parent group voiced similar concerns, saying:

I can't fathom physicians needing to tone it into a different format. That just sounds like a lot of work for whoever is putting that into place. How would you do that? [Parent]

Parent participants speculated about the potential impacts of Open Notes on their child's health care providers. A persistent theme among parents was questioning of the rationale for the Open Notes initiative in general, as opposed to the specific implementation of Open Notes at this hospital or in the pediatric setting. One parent stated bluntly of Open Notes: "I'd be shocked if the doctors really wanted it" (Parent).

Another parent speculated about possible motivations for the Open Notes initiative:

...If physicians are wanting this...is it because they are hoping that parents become more involved?
[Parent]

One parent reflected on her role as a witness of different specialists consulting at her child's bedside and referred to the importance of clinical documentation by all these physicians working in partnership:

[It's] sometimes hard to get the different specialists in the room. They play, in my experience, they play with their brains by writing notes back to each other, or they read each other's notes. [Parent]

Other parents expressed fear that increased transparency of clinical notes would suppress or hinder clinical thinking and dialogue between physicians. These parents were resistant to that potential change:

[D]octors have got to be able to have notes that they can communicate freely so that they can figure out what's going on with some of these kids, because a lot of times they don't know... [Parent]

I think it's really important if we're going to do this that we don't stifle the care and stifle the doctors from doing their jobs. [Parent]

I want doctors to always have the freedom with each other to say, "we don't know, and we're on a

journey”...I want them to have their space to do that, because that’s when the magic happens. [Parent]

Another parent pointed out that interfering with physicians’ communication with other physicians could have downstream negative consequences for the patient and their family: “I wouldn’t want them to hold anything back that may help the child for fear of upsetting me or causing me alarm” (Parent).

Finally, one parent was concerned about the potential impact of Open Notes on the inpatient care workflow:

I don’t want to make their jobs more difficult as doctors, and I don’t want to burden the nurses and other medical staff with all the questions that this could bring up. I mean, I’m very sensitive to the nurses’ time. There are days when the nurses just don’t have enough hours in the day to take care of everybody they’ve got. [Parent]

Medical Jargon

One nurse was concerned about the medical jargon present in the notes and the need to simplify the language for parent viewing based on their experience with the typical educational materials provided to patients:

[E]very teaching material that we give to patients and families goes to our learning center and gets worded to be at...the fifth-grade level or something like that. So even if there’s not medical jargon [in the note], I worry that the language is very far beyond a good portion of families’ reading abilities. [Nurse]

One parent wondered rhetorically whether duplicate notetaking would now be required for 2 physicians to communicate with each other:

If you’ve got to write medical notes at the seventh-grade level, they aren’t medical notes anymore. [N]ow they’ve got to have a different system that they can put their true medical jargon in so that the next specialist knows exactly what they’re looking for. [Parent]

Communication

The theme of communication was mentioned as a benefit in all 5 focus groups; however, it was also noted as a challenge presented by Open Notes. For example, one resident pointed to the complexity induced by the multiple readers and writers involved in note production and the resulting difficulty in interpreting what was meant:

The night team, the two residents on the night taking care of the entire hospital, if now they have to start answering questions that came out about a wording in a note...I can just imagine the increased number of nurses’ pages saying, hey, [the parents] want you to come talk about this note. And that night person isn’t the one who wrote the note. They can’t necessarily say exactly what that person meant at that point in time. [Resident]

These parents also perceived that an intricate balancing act is involved when a writer represents a reader in a note:

[P]roviders noticing family dynamics and commenting on that...could turn into something quite difficult, if there’s a family dynamic that suggests an excessive amount of control or perhaps abuse...having that show up in a note that everyone is seeing...would also be a very delicate circumstance. [Parent]

Another parent commented on the transparency of notes as a communication challenge:

The reason I feel nervous is it changes audience, timing, and delivery all at once. And that’s a lot. [Parent]

Sensitive Content

Among the health care professionals, residents were most vocal about the challenge presented by sensitive content. Specific examples of potentially problematic notes included comments about the family itself:

[I]f there’s things you don’t want the family to know, like you’re considering...they’re neglecting their child, like how are you going to write that here that is friendly? [Resident]

One resident described a potential negative effect on future parent readers who might be frightened by the differential diagnosis process encoded in the note:

[S]ometimes we put...malignancy in the differential diagnosis. And parents, once they read “malignancy,” they don’t care about anything else. Like once somebody hears “cancer,” like that’s the end of their mindset. So, it is going to affect our assessments because we won’t be able to be as clear or as thorough...thinking like how a parent is going to...react to this information. [Resident]

One parent illustrated this phenomenon when they said the following in their focus group:

I think that there’s nothing worse than getting information and feeling like what does that mean? It sounds really ominous. You know, you see a word like “lesion” or “tumor”...and all of a sudden your creative mind runs loose. Weekends and nights are really difficult for things like that. [Parent]

Decreasing Trust

Both residents and nurses said that allowing parents access to notes had the potential to reduce the trust parents placed on their child’s physician. A nurse gave an example of a situation in which a decision had been made to withhold certain information from the family:

I’m just thinking of a specific patient that I had recently...we were concerned about potential conversion disorder, which was a discussion that was had by the medical team but not with the family, because this was a family that was already extremely anxious and extremely...critical of everything that we were doing. And I was already getting questions about...the family being annoyed with the doctors. And obviously, you know, we stick up for our team.

But...[Open Notes is] going to put us in uncomfortable, awkward positions more frequently, I guess?...[W]e already get that sometimes. [Nurse]

A resident spoke about another possible effect of increased transparency on a trusting relationship—the assumption by parents that if one document was open to them, everything could be and should be transparent:

[I]f you do the precedent of...sharing some notes and not others, there's a question of why not? I think that is going to further hurt the relationship in a negative way...why are you hiding? You're not being forthcoming. [Resident]

One parent voiced the same concern when she commented that, counterintuitively, parents might experience decreased trust in their physicians through increased transparency. She argued that they would know that their physician's writing, the documentation of their thought process, was being changed through the expectation of that parent reader:

I would feel like I couldn't trust my physician, because...they were filtering themselves through the hope of this new tool...I want the transparency. I want [the physicians] to go, hey, we could be wrong. And I want the doctors to always have the freedom to go "I'm thinking about this. I could be going down the wrong road." And with this tool, no doctor is going to want to say that in a note. [Parent]

Discussion

Principal Findings

Open Notes advocates have cited compelling reasons to open clinical documentation to patient and caregiver readership in real time. However, research on this question has focused almost exclusively on adult outpatients. This investigation included participants previously underrepresented in the Open Notes research—parents of hospitalized children. All focus groups identified many potential benefits of inpatient Open Notes. These included the enhanced sharing of information between the health care team and absent family members; increasing information for parents to review, thus adding to their knowledge base; providing parents with a sense of structure, enabling them to plan and organize; improving quality assurance for the health care system by involving parents as viewers, commenters, and potential correctors of the record; and illuminating the clinical communication process itself, thus educating and reassuring parents about the care process.

Potential challenges were also voiced. Full transparency of notes carries a risk of reflection: parents might be reading about themselves. In addition, members of all focus groups expressed concern that the process put a burden on health care providers by altering the nature of the note and the note-writing process itself. Parents were worried that these changes would have negative effects on their relationship with their child's physician.

Several benefits discussed in the parent focus group were recurring themes in the Copying Letters research literature. One parent pointed to the capacity of notes to reinforce and recapitulate information that had already been conveyed.

Partridge [15] was originally motivated to copy letters to try and solve this very problem: "Parents and patients often do not remember accurately what doctors have told them." The parents investigated by Richards et al [34] agreed—75% of the parents saying that the letter "reminded me what was said in clinic." Recapitulation was the most frequently mentioned benefit by the 100 parents interviewed by [17], one of whom further validated this perspective when they said:

The things in the letter are helpful, like the dosage of medicines to give. When you are there, it tends to go in one ear and out the other...when you are talking in the hospital, we were worried, so you don't take in what's said, so the letter helps a great deal. [Parent]

Liapi et al [19] also found that their parent respondents liked the summary of the office visit because "it is difficult sometimes to absorb all that the doctor says in the clinic."

Two challenges identified by the participants in this study echoed those voiced by physicians in several Copying Letters studies. The use and readability of medical jargon was mentioned as a potential issue that recurred in nearly all focus groups. Early Copying Letters studies also mentioned jargon as a prospective fear among clinicians who cited this as a reason not to provide copied letters. They believed this, in part, out of concern that parents would not be able to understand medical language and, in part, because avoiding jargon because of a future patient reader would require the physician to "talk down" and degrade communication with other physicians, thus affecting the content and quality of the letters [34,35]. It is interesting that when this theoretical proposition was actually tested by researchers of Copying Letters, parents who had difficulty reading the notes appeared to be in the minority. Cowper and Lenton [17] reported that 96.2% of parents found the language used in the letters "easy to understand." The same result was found years later by Liapi [19]: not one of the 200 parents surveyed experienced an increase in anxiety upon reading their copied letter, and of the 200 parents, only 2 reported any difficulty in understanding medical terminology. Thus, a considerable gap existed between the prospective concerns expressed by clinicians and the actual parent experience.

The same was true for worries about sensitive content. A parent in this study told the other members of their focus group: "You know, you see a word like 'lesion' or 'tumor'...and all of a sudden your creative mind runs loose." As in the case of medical jargon, problems with content recur in the Copying Letters literature as a prospective concern among clinicians; however, like medical jargon, it appears to be a real concern only for a minority of readers. Partridge [15] reported that only 6.8% (9/133) of parents were "seriously upset" by what they read in their copied letters, either because they felt that their parenting was being criticized or because they disagreed with the content. Liapi et al [19] found only one complaint about content: out of 200 parents, 2 "felt that the letter did not accurately describe what they thought was the cause of the child's symptoms." Only 7.8% of the parents surveyed by Amirav et al [18] said that they felt more anxious after reading their child's letter.

The focus groups' ruminations on Open Notes are reminiscent of 2 specific "syndromes" of digital disruption in the wake of EHR implementation: digital deceleration and hypervigilance. Health care systems affected by digital deceleration experience reduced efficiency; digitally hypervigilant individuals are prone to anxiety and overreaction in the face of change [5]. The parents in this study expressed anxiety in remarkably similar ways to health care staff in the same hospital—to the nervous clinicians identified during the Copying Letters initiative in the United Kingdom and the primary and specialist providers surveyed by Richards et al [34]. Like health care professionals, these parents express prospective worries—they “presuffer”—about exposing clinical notes to patients' families before any notes have actually been released. They recognize that the nature of the note itself can be changed through increased transparency and are fearful of the downstream effects of this change. Parental anxieties reveal themselves in comments about note writing: “I can't fathom physicians needing to tone it into a different format,” as one parent puts it; another says “They aren't medical notes anymore if you've got to write medical notes at the seventh-grade level.” The boundary between family and provider could potentially be violated because changing the potential readership changes the actual authorship:

If there's a family dynamic that suggests an excessive amount of control or perhaps abuse...having that show up in a note that everyone is seeing...would...be a very delicate circumstance. [Parent]

They have paid careful attention to the clinical documentation process, from the creation of notes to the reading and sharing of the notes, and acknowledged the importance of notes in the clinicians' workflow. In so doing, these parents repeatedly assume this perspective, as they advocate for the clinical team. These findings highlight the continuing need for clear communication about documentation between parents and providers, including communication about note sharing itself.

Limitations

This study has some limitations. Qualitative data elicited from focus groups are not intended to be generalizable but instead provide the rich context necessary to inform the development of intervention and implementation strategies, in this case, the sharing of inpatient notes. All participants were volunteers; their views may not represent the general pediatric inpatient parent

population. For example, some focus group participants may have had experience with Open Notes in other clinical settings. Whether the anticipated benefits and concerns elicited from the participants in this study will translate into actual outcomes is unknown and an important area for investigation. The impact of sharing notes of other clinicians, such as nurses and physical and occupational therapists, is also a rich area for future research. The benefits, challenges, and impacts of note sharing in the case of adolescent patients are important areas for future investigation.

Conclusions

Sociologist Marc Berg has argued that the medical record is “a force in itself, mediating the relations that act and work through it...The medical record achieves this role through practices of reading and writing” [36]. Until recently, the patient has not participated in medical record viewing. As Hays [37] explains:

Health care professionals have usurped the power to represent patients in the system...and the health record is the primary and most powerful means of accomplishing this...Although the (subjective) voice of the patient is heard, regarding each problem articulated by the nurse, the patient is not a full-fledged member of the fellowship of discourse, is not a reader of the chart and has no responsibility for exchange of the written text.

Thus, providing patients and, in the case of pediatrics, caregivers with access to medical records via patient portals increases the flow of information and, in turn, their ability to participate in the discourse of their care.

At the same time, we must acknowledge the transformative and potentially disruptive nature of this change in the work and dynamic of health care teams. This was a dynamic perceived by parents of pediatric patients themselves. Parents in this study demonstrated not only that they act as monitors and guardians of their children's health but also that they are observers of the clinical processes taking place in the hospital and at their child's bedside. This includes the clinical documentation process, from the creation of notes to the reading and sharing of the notes. Parents acknowledge not only the importance of notes in the clinicians' workflow but also their collaboration with providers as part of the health care team.

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Authors' Contributions

MMK and CAS conducted the conceptualization, data curation, formal analysis, writing review, and editing. Funding acquisition and supervision were done by MMK. Original drafting was done by CAS.

Conflicts of Interest

None declared.

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Abbreviations

EHR: electronic health record

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