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Patient Perspective

From Passive Patient to Engaged Partner: My Journey With Parkinson Disease

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Abstract

This patient narrative by Richard Higgins with Maureen Hennessey describes Richard's journey of learning to live with a chronic and progressive illness. It begins with Richard's diagnosis and shares many of the lessons learned along the way. Richard copes daily with this condition, relying on the support and expertise of his wife and the treatment team he has assembled while also encouragingly drawing on the skills and knowledge gained as a longtime running coach. A clinical commentary is provided at the article's conclusion, drafted by Richard's friend, Maureen Hennessey, PhD, CPCC, CPHQ, offering observations about the relevance of Richard's story to participatory medicine and suggesting pertinent resources for patients and health care professionals.

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KEYWORDS

Parkinson disease; Parkinson disorder; parkinsonism; movement disorder; physical exercise; aerobic exercise; physical activity; running; race-walking; patient self-care; patient self-advocacy; patient self-efficacy; coaching; health coaching; care management; patient engagement; shared decision making; participatory medicine; patient-centered care; patient-centered outcomes; caregiver support; positive psychology; Parkinson disease quality measures; quality management

Patient Essay by Richard Higgins

My Diagnosis

In the spring of 2009, after falling down the stairs several times within a week, I knew something was wrong. I needed to find out what it was. I received an initial diagnosis of Parkinson disease (PD) and was referred to a movement disorder specialist for confirmation. They said it was not PD and sent me back to the first neurologist. He then gave me a tentative diagnosis of a parkinsonism.

Next, I was sent to a Movement Disorder Center where, more than 2 years and 4 doctors into this journey, I was diagnosed with PD. The parkinsonism that was earlier suspected typically progresses much faster and does not respond as well to current treatments, so my wife and I were relieved by the diagnosis. Then, the question became, what to do about it?

PD includes an array of symptoms beyond the tremors, shaking, and shuffling steps that are familiar to the general public. Symptoms vary in sequence and severity for every person. These include stiffness and rigidity, inability to reflect moods on your face, reduced fine motor coordination, disrupted sleep, decreased sense of smell, balance issues, mild cognitive impairment, depression, poorer/smaller handwriting, delayed reaction time, and difficulty swallowing. Like many chronic conditions, medications have helped with some symptoms, but do not cure or retard the progress of this illness.

Prior to my diagnosis, I had always taken a passive role in my health care. Not being a doctor or clinician, I have been accustomed to doing what physicians tell me to do or, in less serious matters, following suggestions in various fitness articles. Yet, due to the symptom complexity and variability of PD, it became clear that no single doctor could manage all of my care or could know everything I needed to do to manage my condition.

Becoming an Engaged Participant in My Care

It was good news when my neurologist mentioned that intensive aerobic exercise has been shown to slow progression of PD, since I enjoy running and race-walking. My experience as a cross-country and track and field coach helped me tap into my knowledge of goal-setting, physical activity, and planning workouts to help not only my students but also myself.

In the ensuing years, I tailored my training plan to try to minimize the impact of PD for as long as possible. I transitioned from being serious about my workouts, especially my cardio sessions, to pushing myself to a nearly fanatical degree. I was able to incorporate various exercises and techniques from specialists I consulted (such as physical therapists, chiropractors, and strength and conditioning coaches) into my routines. I created a plan that I could modify as my condition changed and as I learned new things. This plan now includes a combination of aerobic and cardio intensity, balance exercises, core conditioning, general body strength, and fine motor coordination. As I learn and my efforts evolve, several important concepts have emerged:

1. No matter the stage of PD, some type of exercise is beneficial. Muscles atrophy quickly if not used. Muscles that are used degenerate more slowly.
2. Be involved in, and committed to, your training plan. While sometimes it can be a struggle even to do the routines you enjoy, planning your workouts helps you “buy in” to them more completely.
3. Incorporate enjoyable activities into your plan. It’s easier to stay with a routine—particularly one you need to do long-term—the more you enjoy it. This can also provide some feeling of control with an unpredictable disease.
4. Begin slowly. Increase the amount, types, and intensity of exercises just a little each week. Remember: always discuss any plans for physical activity with your physician(s).
5. Learn what exercises are within your ability and which can be beneficial. With any chronic condition, one exercise routine does not fit all (especially with PD). Establish baselines, set goals, and measure progress.
6. Take advice from a variety of professionals. Over time, professionals such as occupational therapists, psychologists, athletic trainers, and aging specialists may become part of your treatment team. Candor about your condition, including your symptoms, reaction to drugs, and amount you exercise, helps your clinicians to better help you. It may help if you provide them with information before appointments so they are prepared, but not all clinicians will be able to work that way.
7. Include caregivers early. I also think of my wife as my “care-partner.” Caregivers or care-partners can help you solve problems in adapting to living with PD, be a coadvocate for you, and provide information to your clinicians that you may forget. It will also make it much easier for them to transition into this role.

Learning to Ask for Help: Lessons in Grace and Humility

This last point—knowing the need for and accepting a caregiver—has been critical in handling the struggle with PD. As my symptoms gradually worsen, I have had to accept that there are some things I just can’t do safely. Tasks to avoid, like washing sharp knives or crystal glasses, dawned on me long after becoming obvious to my wife. Unfortunately, it took much longer to realize that my reluctance to accept her help only increased her burden. Not communicating to her my symptoms, frustrations, and needs (which I am still trying to perfect) can cause her to expend more energy and time. As the PD continues to progress, she has gradually assumed more of a care-partner role. There is added urgency now for me to talk with her about my successes, failures, and plans. When she knows what is happening and what I am doing, she is better able to help me coordinate my care, rather than having to guess what I really want or need.

Surprising myself, I decided to give up driving. After developing a delayed reaction in my right foot and leg, I realized that having any hesitancy while operating a vehicle could prove disastrous. Unfortunately, this also led to the realization that I needed help from other people to get where I had to go. I resented this loss of freedom, as well as having to ask for help, accepting it when it was offered, and being grateful for it. I have had no difficulty finding people to drive me and am indebted to my wife and daughter, as well as to fellow coaches and parents of cross-county team members. But it has been difficult admitting (even to myself) that I need assistance. This has improved over the last several years but is always hard. When I decided to approach this challenge the way I approached my exercise routine, I began setting goals for communicating my needs and expressing appreciation for the support. This has helped me maximize my independence while also accepting my limitations with grace.

More Lessons Learned: The Journey is Always Evolving

I’m now in the ninth year of my journey. There have been many joyous milestones (graduations, a son’s wedding, a daughter’s wedding, and the birth of grandchildren) that make me realize how important it is to do all I can in this progressive fight. There have been exciting times watching students win cross-country and track championships and going on wonderful trips with my wife. However, I also must learn to deal with setbacks. Any plan, no matter how well thought out, can go awry at times. One winter, I was not prepared for just such a setback. A required minor surgery prevented me from exercising for nearly 4 months. Not only did I lose a lot of conditioning, but also a routine I depended on psychologically. I experienced some changes in physical symptoms, some loss of cognitive ability, and signs of depression. When I was able to exercise again, I also had to work on my mental and emotional conditioning. I have come back somewhat, but not yet to presurgery levels.

I have learned a great deal throughout this journey. First and foremost, to remain as active as possible, so I can enjoy more great moments. I have learned there always will be unavoidable challenges. For example, during the last few months, I have undergone additional testing. This has revealed several atypical indicators and symptoms, suggesting the possibility that my PD condition may present atypically or could perhaps be another parkinsonism. Depending on what is ultimately learned, my treatment and self-care may require more adjustments.

While there are many advisors who can help during those times, I have learned that my caregiver(s) and I are the people who can best manage this team. I have learned to be involved in my care and graciously allow others to help when needed while continuing to do the things I can do. I am still learning these things, and it is not always easy. Planning with PD is tough, as my symptoms change from day-to-day and even throughout the day. It helps to prioritize exercise and to focus on the importance of enjoying life while coping with this difficult condition. Although there is no cure for PD yet, I continue to be hopeful about the many new treatments to help manage it. Overall, staying active through exercise and being involved in my care helps me be more positive about today and the days to come.

Invited Clinician Comment by Maureen Hennessey

This narrative describes a patient's journey of learning to live with a chronic and progressive illness. It begins with the diagnosis, moves through patient participation, and emerges with many lessons learned along the way. Coping daily with this condition, he relies on the support and expertise of a spouse and other family members, and the treatment team assembled, while encouragingly drawing on the skills and knowledge gained as a longtime running coach. His story exemplifies a key concept in participatory medicine: all of us must be participants in and contribute to our care, to the extent of our preferences and abilities. We can all admire his courage, candor, and tenacious self-care, exemplified by his thoughtful approach of seeking and accepting assistance and support. As clinicians, care managers, health coaches, and health quality experts, we can learn from descriptions of how patients and caregivers collaborate and partner with each other and with health care professionals to seek and use information resulting in well-informed personal choices. Patient goal-setting, problem-solving, and positive psychology are instructive, particularly as patients strive to enjoy life and help others while hoping for better treatments to manage their conditions.

At least 10 million individuals worldwide, including more than 1 million Americans, live with PD [1]. For additional patient and caregiver education and resources, and information about finding a cure, you can visit the American Parkinson Disease Association [1] website and the Michael J. Fox Foundation for Parkinson's Research [2] website. Information about quality measures pertinent to the treatment of PD (including exercise, mood, and rehabilitation) may be found in "Quality improvement in neurology: Parkinson disease update quality measurement set" on the American Academy of Neurology website [3].

Conflicts of Interest

None declared.

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Abbreviations

PD: Parkinson disease

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Viewpoint

Experiencing Positive Health, as a Family, While Living With a Rare Complex Disease: Bringing Participatory Medicine Through Collaborative Decision Making Into the Real World

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Abstract

Physician–patient collaboration was recognized as a critical core of participatory medicine more than a century ago. However, the subsequent focus on scientific research to enable cures and increased dominance of physicians in health care subordinated patients to a passive role. This paternalistic model weakened in the past 50 years—as women, minorities, and the disabled achieved greater rights, and as incurable chronic diseases and unrelieved pain disorders became more prevalent—promoting a more equitable role for physicians and patients. By 2000, a *shared decision-making model* became the pinnacle for clinical decisions, despite a dearth of data on health outcomes, or the model's reliance on single patient or solo practitioner studies, or evidence that no single model could fit all clinical situations. We report about a young woman with intractable epilepsy due to a congenital brain malformation whose family and medical specialists used a *collaborative decision-making approach*. This model positioned the health professionals as supporters of the proactive family, and enabled them all to explore and co-create knowledge beyond the clinical realm. Together, they involved other members of the community in the decisions, while harnessing diverse relationships to allow all family members to achieve positive levels of health, despite the resistance of the seizures to medical treatment and the incurable nature of the underlying disease.

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collaboration; shared decision making; patient–physician relationship; communication; partnership; participatory medicine

Introduction

Collaboration between physicians and patients in a mutual relationship—the core attribute of participatory medicine—was first documented in the late 19th century [1]. Bertha Pappenheim (also known as Anna O.) and Dr Josef Breuer, her physician and Freud's mentor, discovered the therapeutic power of a collaborative partnership in the 1880s while listening to and learning from each other [2]. In the late 1950s, the *mutual participation model* was relegated to psychoanalysis and psychology. Traditional medicine favored models that had existed since Ancient Egypt, which placed physicians as the dominant members of the relationship, and patients mostly as passive or inactive beings. These *paternalistic*, *priestly*, or *passive* models [3-5] pervaded 20th century medicine, driven by a hierarchical view of knowledge, with physicians at the apex and patients/families at the base, a reductionist *chemical–mechanical* view of people, a pathophysiological approach to illness, and a belief that science could conquer disease and even defeat death [6].

As most acute conditions (eg, infections, diabetic coma, appendicitis) became curable, chronic incurable diseases dominated, leading to models based on *doing things to patients* [7]. This approach to chronic disease is rife with limitations, and in some cases (eg, chronic pain and psychiatric disorders), modern medical approaches may do more harm than good. By the early 21st century, a *shared decision-making model* had gained ascendance as *the pinnacle* for clinical decisions, particularly within the context of evidence-based medicine and patient-centered care [8-11]. Even though there are multiple ways to conceptualize it, in essence, shared decision making incorporates at least two participants—typically 1 physician and 1 patient—who examine information about different options to manage a condition, taking steps to build a consensus and to agree about which one to implement [12].

The shared decision-making model echoes and builds on precursors from the mid-1900s, which failed to cross from theory into clinical practice. Current approaches reflect the reaction against paternalism in the physician–patient relationship. They received names such as *mutual participation* or *collegial* models [3,7], and were also regarded as alternatives to another model, which considers physicians as the main source of facts and synthetic advice, for patients to weigh relative values and make the diagnostic or therapeutic decisions. These have been labeled *consumerist*, *informative*, *informed*, *autonomous*, or *engineering* models [3,7,13-16]. Despite being touted as the ultimate model, several systematic reviews of shared decision making reveal a dearth of supportive evidence, leaving their impact on empirical health outcomes uncertain [10,17-23].

Others observed that most research on shared decision making does not match clinical reality, because studies focus on a single patient with a solo practitioner. Instead, the real-life situations employ the model with patients who do not want to make decisions alone, preferring their loved ones to be involved or take charge in making critical decisions, and with multiple specialists participating in their care [24]. Besides, patient preferences for a shared decision-making model vary across studies according to their date of completion, as well as the selected population and the measurement tools used [25]. Further, the sheer diversity of models of relationship might indicate that the needs and preferences of patients and clinicians differ, and that existing models are components of a menu from which to choose, rather than single, fixed options to use during their interactions [26].

Our case illustrates how a new model, *collaborative decision making*, enabled a family and a group of involved health professionals to overcome all of the aforesaid limitations. This new approach, which was proposed in this journal in 2010 as an invitation to those involved in participatory medicine to consider a shift from the *shared* model, is presented here as an option to enrich, rather than to replace or displace, all other options, as it could foster a stronger partnership among patients, loved ones, and professionals, encouraging them to engage in a process with the common goal of creating a plan of action aimed at improving health [27].

The description follows the parameters that reflect the range of interests of those involved in participatory medicine, and underscores the desire of an entire family that leveraged this model to find solutions not offered by leading institutions, and to bring their experiences to other patients and health professionals who could learn from it.

Case Presentation

The People

Silvana was a 14-year-old woman when she was diagnosed with subcortical band heterotopia in 2011, following a seizure during a flight. This rare condition results from millions of neurons that do not migrate properly during development, creating a brain with dense bands below the cerebral cortex, where there should only be white matter fibers connecting neurons [28]. This explained the mild learning impairment that Silvana experienced throughout her life and the drug-resistant focal epilepsy that was progressively worsening for 5 years. She had 4 different seizures types, occurring at least once per week each but some up to 30/day, and lasting 4-150 seconds. Typical seizures included a fixed stare, shaking, or trembling of one hand, without loss of awareness, followed by fatigue. Every few months, she would have a drop episode. Despite these challenges, Silvana remained a cheerful young woman, keen to be offered tasks to complete, and eager to engage in artistic pursuits, especially photography and painting. Her main concern was, consistently, not to be left alone in an enclosed environment, such as an elevator, because of her fear of injury as a result of a fall.

The severity of Silvana’s condition disrupted her family’s life, with her father Ricardo most affected. He was frustrated by the trial-and-error approach to the frequent changes in the dose and combination of antiseizure medications by the multiple physicians involved in her case, despite understanding the dearth of scientific evidence supporting any option over the others. Using the skills and attitudes that had enabled him to become a successful entrepreneur, such frustration was transformed into a relentless urge to become an expert on band heterotopias and to perform online searches, almost compulsively, seeking to find a silver bullet that could have been missed by all of the specialists involved in his daughter’s care. He also joined groups of parents on social media, hoping to find and benefit from additional insights from the field. The frustration associated with the failure to find an effective treatment for the seizures morphed into exhaustion and anxiety so intense that he required support from psychologists and psychiatrists, with little benefit. An additional source of distress was the regret produced by the realization that he would feel much better whenever Silvana was out of his sight, especially in a different city.

Silvana’s mother, Denise, faced different challenges. She accepted the problem’s incurable nature and its complexity, which meant no doctor had sufficient data to guide therapy accurately, and sought to reduce its social impact. She fought the stigmatization by the family’s relatives and friends as well as potential rejection by peers, and the interruption of Silvana's high-school studies. Given the conservative city in which they lived, the family decided not to disclose the underlying neurological condition to the school until the last year of studies, or to people outside the inner family circle. Instead, Silvana was diagnosed with a learning impairment.

Leon, Silvana's only sibling, became very supportive of his sister, while maintaining his high academic level of school performance. Even as a young child accompanying his sister to medical visits, he was able to ask pertinent questions about *why* this happened to his sister's brain, underscoring that the common sense questions of a child are among the most important questions that physicians should try to answer.

New Knowledge Creation Through Collaboration of Researchers and Patients, as Individuals and as Groups

By 2016, Ricardo's relentless efforts to find effective medicines to control Silvana's seizures proved fruitless. As this was making his distress overwhelming, a physician friend of the family (Miriam Tabacinic), who was aware of the collaborative decision-making model, suggested to contact its lead author (Alex Jadad), whom she knew since their postgraduate training years back in the 1980s.

Given that they were located in different regions of the continent, Ricardo and Denise held an initial virtual conversation with Alex, during which the latter explained the model, emphasizing the need for a shift from a focus on the fight against the disease to one devoted to the enjoyment of health, and from Silvana to Ricardo as the person most in need of support.

Ricardo's level of distress was extremely high and reducing it became the first priority. Because of his compulsive desire to cure Silvana's seizures and the large swaths of time he was spending searching the biomedical literature and consulting specialists in different regions of the world, it was agreed that a formal synthesis of the literature on treatment-resistant epilepsy would be conducted, and that the leading authors would be invited to join a panel to discuss Silvana's case and the best course of action.

The synthesis, which included papers indexed by MEDLINE and EMBASE from January 2015 to June 2016, was complemented by screening of all of the citations of relevant articles, and a forward search, using Google Scholar.

This systematically individualized effort to search, screen, and distill the peer-reviewed and gray literature revealed many options with a high probability of success still available to Silvana, including cannabidiol and other cannabinoids; conventional, first-line antiepileptic drugs; ketogenic or modified Atkins diets; noninvasive neurostimulation methods; experimental drugs; vagus nerve stimulation; or corpus callosotomy.

Health Professionals and Health-Related Institutions

The relevant articles identified potential experts who were invited to become panel members to hold an in-depth discussion about which options to pursue (see Acknowledgments). They included Orrin Devinsky (Panel Chair, who also was Silvana's current treating physician) and Annapurna (Ann) Poduri, another physician in Silvana's team who managed her as a teenager and was highly trusted by her family.

The group acknowledged that a diverse panel of experts provided an opportunity for new insights to emerge to maximize Silvana's health, while reassuring Ricardo and the family about the robustness of the recommendations. The panel held one whole-group session, chaired by Orrin, using a digital videoconferencing platform, which was followed by on-demand ad hoc email exchanges. After multiple interactions, invasive options were unacceptable to the family. Instead, it was decided to try a modified Atkins diet coupled with different combinations of conventional pharmacological interventions, leaving transcranial stimulation and experimental drugs to be considered at a later stage.

As Silvana was now an adult, it was agreed that Orrin would act as the main treating physician, working closely with Ann, to ensure continuity of care.

Contextual Determinants

The rigor of the panel, the commitment of its members, and the open and comprehensive way in which Ricardo's questions were addressed enabled a major shift. The family's near-exclusive focus on the illness and its symptoms was broadened and redirected to a more constructive emphasis on health, acknowledging that it is much more than the absence of disease. They accepted a conceptualization that considers health as the ability of individuals and communities to adapt and manage the physical, mental, or social challenges faced throughout life [29]. This facilitated a much more effective and natural alignment between the family's goals, the views of the experts, and the collaborative decision-making model to develop *an optimal action plan to improve health* [27].

This shift to a health-focused approach to Silvana's life with intractable seizures enabled a transition from finding a cure or complete seizure control to achieving maximum levels of adaptation and self-management of an incurable condition through the activities illustrated in Table 1. Throughout the process, they agreed to monitor their levels of self-reported health by asking themselves the following question: "In general, would you say that your health is poor, fair, good, very good, or excellent?" Answering *poor* or *fair* represented negative health, whereas *good*, *very good*, or *excellent* was regarded as reflecting positive health.

Initially, Ricardo's self-reports were consistently negative; Silvana's and Leon's were consistently positive; and Denise's fluctuated, depending on whether she spent more time with Ricardo or their children.

The intention was to achieve positive health self-ratings for all family members for at least six months, to consider the approach successful and worth sharing with other families and health professionals. Figure 1 summarizes the journey.

Table 1. Efforts to generate maximum levels of adaptation and self-management within the context of health-focused collaborative decision making

Goals	Health domains		
	Physical	Mental	Social
Adaptation	Companionship in enclosed spaces and outdoors to prevent injuries	Recognition of the incurable nature of the disease	Adapted high-school curriculum On-site tutor to assist with academic tasks
Self-management	Optimal adherence to medication intake	Family-focused counseling, yoga, and meditation	Initiation of a small business with a close family friend

Figure 1. Key stages of the collaborative decision-making process within the context of participatory medicine.

1\.

Onset

Seizure during a flight when Silvana was 14 years old, followed by drug-resistant focal epilepsy that became progressively worse during the following five years.

2\.

Diagnosis

Subcortical band heterotopia.

3\.

Trial-and-error approach to therapy

Specialists at multiple institutions had prescribed multiple medications, including Oxcarbazepine, stiripentol, levonorgestrel and ethinyl estradiol, Levetiracetam and Vitamin B6, without clear benefit.

4\.

Family frustration

Resistance to treatment caused major disruption of the entire family's life. Ricardo, the father, experienced intense frustration and distress that required psychological and psychiatric support.

5\.

Shift to a collaborative decision-making approach

In 2016, with support from Drs Jadad and Tabacinic, the family agreed to focus on experiencing positive health, and to reduce emphasis on fighting the disease.

6\.

Formal synthesis of the literature

A comprehensive search of MEDLINE and EMBASE from January 2015 to June 2016 was complemented with a forward search of Google Scholar. All of the citations of relevant articles were screened and analyzed.

7\.

Panel of experts

Key studies were used to identify and invite experts to have in-depth discussions about unusual therapeutic options, as part of a panel. Dr Orrin Devinsky acted as Chair and Dr Jadad as facilitator.
(All members are listed below).

8\.

Alignment of goals

The panel reassured the family and promoted the development of "an optimal action plan to improve health", which is the overarching objective of the collaborative decision-making model.

9\.

Adaptation and self-management

Considering health as an ability, the family transitioned their efforts from trying to control the epileptic episodes completely to achieving maximum levels of adaptation and self-management of an incurable condition.

10\.

Monitoring of self-reported health

The family assessed their levels of self-reported health regularly, by asking themselves: In general, would you say that your health is poor, fair, good, very good, or excellent? Answering good, very good, or excellent was regarded as reflecting positive health.

11\.

Challenge management in all domains

To achieve positive levels of self-rated health for all members of the family, specific adaptation and self-management goals were pursued in the physical, mental, and social domains.

12\.

Sustained health improvement

By February 2020, all family members reported positive levels of health for more than 6 months. Silvana had seizure-free periods for up to 3 weeks, without convulsive seizures for more than a year.

Panel of Experts

Orrin Devinsky (Chair, neurologist, and current main treating physician), **Annapurna Poduri** (neurologist, main treating physician during Silvana's teenage years), **Miriam Tabacinic** (physician and trusted family advisor), **Alejandro Jadad** (physician, expert in evidence-based and collaborative decision-making), **Eric Kossoff** (pediatric neurologist, with interest in nonpharmacological interventions), **Lieven Lagae** (neurologist with interest in drug-resistant epilepsy), and **Fergus Rugg-Gunnfor** (neurologist with interest in new and non-pharmacological interventions for epilepsy).

From the physical perspective, the diet added little value in seizure control despite sustained strict adherence, and medication reduced seizure frequency and duration without eliminating them. Therefore, the collective objective switched to reducing bodily injuries while more drugs were tried following a systematic trial-and-error approach.

From the mental point of view, the panel contributed significantly to Ricardo's acceptance that Silvana's condition was incurable and that she was receiving the best available therapies. Subsequently, the family engaged in individual yoga and meditation training, and group counseling to identify and set boundaries, which facilitated their collective adaptation to living with an intractable chronic condition. Silvana was encouraged and supported to build self-confidence, and to develop new ways to manage fears and strategies to respond to frustrating situations calmly. The entire family engaged in activities to reduce the reinforcing of Silvana's sick role and to explore spiritual practices. The latter were particularly relevant for Ricardo, who decided to reconnect to his religious roots, finding solace and new sources of strength to deal with stress and to eradicate his guilt.

Socially, Silvana completed her high-school bilingual education with an adapted curriculum and support from an on-site tutor. Based on Silvana's enjoyment of tasks requiring attention to detail, her family supported the creation of a small business focused on the manufacture and commercialization of one-of-a-kind fashion accessories with a close family friend, which proved that it would be possible for her to make a living doing something she likes.

Soon, all of these changes became part of their normal daily living, and their self-reported health status had stabilized at positive levels for all family members (Figure 2). Silvana summarized the situation by stating, "I feel better than I did before now that I have changed my habits. Yoga helps me concentrate on my breathing, and increases my awareness level. My family helps me by providing me with loving support and balance in my life."

Figure 2. Denise, Silvana, Ricardo, and León Caridi in 2020.

By February 2020, Silvana was taking lacosamide (400 mg/day), clobazam (40 mg/day), and vigabatrin (1750 mg/day), enjoying a significant and sustained reduction in her seizures. The most severe seizures caused head drop with impaired awareness for up to 30 seconds. She could go for up to 3 weeks seizure free.

Anxiety and stress remained triggers, and when she reduced her daily yoga activity, her seizures would worsen.

By the time this article was submitted, Silvana was working with a cousin in her photography and graphic design company, designing a family cookbook. At that point, she had been free of convulsive seizures for more than a year.

Discussion

This case illustrates how collaborative decision making could enable a family and their health professionals to use different relationship models, to explore therapeutic options within the broad domain of traditional and nontraditional medicine. The panel enabled Ricardo to benefit from the informative model by asking the participating specialists to answer his questions until he was reassured that the family's decisions were based on the best knowledge. Once this was achieved, the panel promoted two-way knowledge exchange, shifting to a shared decision-making model, and focused on identifying the best course of action. Once there was agreement around the best course to follow, the family comfortably switched to a slightly paternalistic relationship, especially for diet implementation and systematic medication changes.

Conceptualizing health as the ability to adapt and self-manage the physical, mental, and social challenges created by the subcortical band heterotopias added value in fundamental ways. First, the family shifted years of emphasis on the disease to concentrating on achieving optimal levels of health per se. Part of this process was giving themselves permission to deviate from the *cure at all cost* paradigm. Second, this conceptualization helped the family recognize that all of them—not just Silvana—needed support to enhance their capacity to adapt to her intractable epilepsy. Lastly, it showcased how team effort can make positive health possible even in the presence of chronic, incurable diseases, opening new avenues for patients and clinicians to harness the power of collaboration as the essence of participatory medicine.

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Conflicts of Interest

None declared.

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Original Paper

Non-Hispanic White Mothers' Willingness to Share Personal Health Data With Researchers: Survey Results From an Opt-in Panel

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Abstract

Background: Advances in information communication technology provide researchers with the opportunity to access and collect continuous and granular data from enrolled participants. However, recruiting study participants who are willing to disclose their health data has been challenging for researchers. These challenges can be related to socioeconomic status, the source of data, and privacy concerns about sharing health information, which affect data-sharing behaviors.

Objective: This study aimed to assess healthy non-Hispanic white mothers' attitudes in five areas: motivation to share data, concern with data use, desire to keep health information anonymous, use of patient portal and willingness to share anonymous data with researchers.

Methods: This cross-sectional study was conducted on 622 healthy non-Hispanic white mothers raising healthy children. From a Web-based survey with 51 questions, we selected 15 questions for further analysis. These questions focused on attitudes and beliefs toward data sharing, internet use, interest in future research, and sociodemographic and health questions about mothers and their children. Data analysis was performed using multivariate logistic regressions to investigate the factors that influence mothers' willingness to share their personal health data, their utilization of a patient portal, and their interests in keeping their health information anonymous.

Results: The results of the study showed that the majority of mothers surveyed wanted to keep their data anonymous (440/622, 70.7%) and use patient portals (394/622, 63.3%) and were willing to share their data from Web-based surveys (509/622, 81.8%) and from mobile phones (423/622, 68.0%). However, 36.0% (224/622) and 40.5% (252/622) of mothers were less willing to share their medical record data and their locations with researchers, respectively. We found that the utilization of patient portals, their attitude toward keeping data anonymous, and their willingness to share different data sources were dependent on the mothers' health care provider status, their motivation, and their privacy concerns. Mothers' concerns about the misuse of personal health information had a negative impact on their willingness to share sensitive data (ie, electronic medical record: adjusted odds ratio [aOR] 0.43, 95% CI 0.25-0.73; GPS: aOR 0.4, 95% CI 0.27-0.60). In contrast, mothers' motivation to share their data had a positive impact on disclosing their data via Web-based surveys (aOR 5.94, 95% CI 3.15-11.2), apps and devices designed for health (aOR 5.3, 95% CI 2.32-12.1), and a patient portal (aOR 4.3, 95% CI 2.06-8.99).

Conclusions: The findings of this study suggest that mothers' privacy concerns affect their decisions to share sensitive data. However, mothers' access to the internet and the utilization of patient portals did not have a significant effect on their willingness to disclose their medical record data. Finally, researchers can use our findings to better address their study subjects concerns and gain their subjects trust to disclose data.

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KEYWORDS

surveys and questionnaires; confidentiality; electronic health records; fitness trackers; mobile applications; logistic models

Introduction

Background

Advances in information communication, electronic health (eHealth), and mobile health technologies are increasingly used to access and collect personal health data. These have contributed to the expansion of research in health care and public health. The eHealth apps can be Web-based or mobile apps that include a range of features, such as tracking changes in health behaviors, chronic care management by health professionals or the patients themselves, or location tracking. In this regard, many eHealth studies have focused on adult women, and specifically mothers, who have often been used as proxy respondents for studying the diverse health care issues of their families [1]. Moreover, there has been an increasing interest among commercial organizations and researchers in recruiting mothers to gain insight into their behaviors while using the internet [2,3]. In 2014, 85% of white women in the United States used the internet [4]. In an eMarketer report [5], mothers, if that is true, more often requested immediate and continuous access to the internet, compared with total female consumers in general (53.9% vs 44.3%, respectively). Women, particularly mothers, tend to use the internet to look for health and medical information, get support from different sources, and search for other information needs [6,7]. Although research in the area of data sharing is increasing, many questions remain to be answered that mostly pertain to privacy and data-sharing preferences.

Privacy and Data Disclosure

Privacy and data disclosure are considered serious challenges for health care researchers, and patients are hesitant about sharing their health data as they may expect a loss of privacy while sharing their personal information. This issue encourages patients to retain control over their personal information and disclosure of their data. In fact, patients undergo a cost-benefit analysis to assess different factors that influence their preferences and decisions to share their data, which is known as the privacy calculus [8-10].

Although a substantial body of literature has examined individuals' willingness to share their health data, most of these studies have focused on health information exchanges in the general population [11-19]. These findings have created interest in how to solicit information from patients and maximize their participation in research studies. Most studies have found that data security and privacy preferences shape consumers' attitudes toward health information exchanges [11,12,14-16,18-21]. As reported, consumers perceive health information exchanges to confer benefits, such as better coordination of care [18] and improvement of health care quality [14]. Accordingly, researchers have considered privacy control in the context of research to enhance the engagement of individuals and establish trust with the study participants.

Objective

An attempt to find a solution to greater privacy control has increased the number of studies addressing privacy on the basis of differentiation between sensitive and less sensitive personal health data. In this regard, consumers have more choices to share their personal data with whom they feel more comfortable [22]. Therefore, innovative systems are being developed to give individuals more power to determine the researchers who are allowed to access their data and the type of data they are willing to share [23]. However, the granularity of data necessary for this level of privacy increases the burden on stakeholders (ie, researchers) and a priori market research, as well as market segmentation, can facilitate the assessment of consumers' willingness to share their data. Accordingly, a number of studies have sought to gauge how sensitive consumers are about sharing their data and the amount of data they are willing to disclose. As a result, this study aimed to assess healthy white mothers' attitudes toward the anonymity of their health information and their utilization of the Web-based patient portals and willingness to share different data sources with researchers.

Methods

Study Design

A total of 622 women were randomly selected from a commercial opt-in panel with several million US members. The participants were non-Hispanic white women with children from the Centers for Disease Control and Prevention–funded study on the care burden of mothers of children with autism spectrum disorder (ASD), implemented collaboratively by the Department of Health Management and Informatics of the University of Missouri School of Medicine and the Kennedy Krieger Institute's Interactive Autism Network (IAN). The study compared care burdens and associated factors between a US representative sample of mothers and the sample of mothers of children with ASD from the IAN registry. As the proportion of nonwhite mothers in the IAN registry was very small, the design required the selection of a comparative US sample of white women raising children without disabilities. According to the sampling method of the original study, the samples in this study were representative of white mothers aged 25 years or older living in an urban area. The majority of the investigated individuals had an educational level of college (4 years) and were employed or self-employed.

Survey Questionnaire

The research tool was a survey questionnaire with different domains related to attitudes and beliefs, trust in data sharing, data sharing through mobile phone apps and devices, internet use, and interest in future research, as well as questions related to caregivers and their children. The survey questionnaire consisted of 51 questions, of which 15 were selected for further analysis. The questions selected were related to attitudes and beliefs about data sharing, internet use, and interest in future research, sociodemographic information, and health questions about caregivers and their children.

Outcome Variables

On the basis a review of empirical studies [7,24,25], mothers were assessed based on their preference for keeping their health data anonymous. As reported in other studies, patients are more willing to disclose their data when the data are anonymously shared with researchers [26]. In this regard, respondents were asked to answer the following question: “How important is it to you that your personal health information is kept anonymous?” To answer the questions, the respondents could choose from the following four choices: *extremely important*, *very important*, *somewhat important*, and *not at all important*. The *extremely important* and *very important* answers were grouped into an *extremely important* category for analysis.

Similarly, mothers’ familiarity with patient portals was investigated in this study, as familiarity with the source of health care data may influence patients’ willingness to share their data. For instance, a study conducted by the United States Department of Veterans Affairs indicated that veterans had a higher level of willingness to share their health information when they gained a higher level of familiarity with the utilization of a Web-based portal [24]. Therefore, it could be suggested that when the majority of the mothers are familiar with the patient portal, they will be more inclined to share their medical records with researchers [24]. Accordingly, respondents were asked about their familiarity with the patient portal and frequency of its use. The answer provided by the investigated participants could be one of the following alternatives: *never heard of it*; *heard of it but never used it*; *yes, once a month*; and *yes, more than once a month*. The answers were then dichotomized into two categories *never used it* (combining never heard of it and heard of it but never used it) and *have used it more than once* (combining *yes, once a month* and *yes, more than once a month*).

The respondents were also required to answer the question, “Which type of health-related data are you willing to share anonymously with researchers?” They could choose from the following choices: *data from medical records*, *data you provide directly by completing an online survey*, *data you entered into a health-related mobile app or device*, *data collected automatically by mobile app or device*, and *GPS location data from a mobile device*. Answers to the following choices *data you enter into a health-related mobile app or device*, *data that is collected automatically by mobile app or device* and *data entered into or collected through mobile app or device* were considered as a positive category for sharing data for analysis.

Independent Variables

To address respondents’ privacy calculus [26], two questions were included to measure respondents’ concerns and motivations for sharing their health data. The respondents were asked, “How concerned are you that your health record might be…”, to which they could select one of the following choices: *used to deny me health care benefits*, *used to limit my job opportunities*, *used without my knowledge*, and *stolen by unknown individuals or companies*. They could rate these choices as *very concerned*, *somewhat concerned*, *only a little concerned*, *not at all concerned*. If the answers provided to all the choices were *very concerned*, the score would be 4, and the response *not at all concerned* for all choices resulted in a total score of 16. Respondents with the total score <8 were categorized as *very concerned*, those with scores ranging from 8 to 11 as *somewhat concerned*, and individuals with scores >11 as *less concerned*.

Similarly, to assess mothers’ motivation to share data with researchers, the participants were asked, “What is your motivation for sharing your health information?” The mothers could select any of the following choices: *benefit future patients*, *contribute to science and research*, *trust in researcher organization, and a desire to contribute to the research they are doing*, *establish a good relationship with researcher organization*, and *other*. The obtained motivation score was the sum of the selected answer choices ranging from 0 to 5. A score of 0 meant that the respondent did not select any choices, and we categorized this respondent as *not motivated*. On the other hand, the score of 5 was indicative of mothers who selected all the answer choices, so they were *highly motivated*. In the next step, the motivation scores were divided into three levels. The individuals with motivation scores ≥1 were grouped into *less motivated*, whereas those with motivation scores between 1 and 3 were grouped into *somewhat motivated*, and motivation scores >3 were *very motivated*.

We dichotomized the variables of age (18-49 and >50 years), education (less than 4 years of college and 4 or more years of college), occupational status (self-employed and other occupational status), income level (household income ≤US $74,999 and ≥US $75,000), marital status (married and other status), age of the child/young adult (≤14 years and ≥15 years), number of children (1 child and more than 1 child), health status of the youngest child/adult, and mothers’ health status in general (excellent to good and fair to poor) based on their frequency distribution.

Data Analysis

Data analysis was conducted using SAS (version 9.3). Moreover, the frequency analysis was carried out to describe the demographic characteristics of the surveyed mothers, their data-sharing preferences, and their privacy concerns [27]. Multivariate logistic regression was also conducted to investigate the associations of selected outcomes and a set of independent variables. In the models, the dependent variables were *desire to keep personal health information anonymous*, *use of patient portal*, and *willingness to share anonymous data with* *researchers regarding medical records, Web-based survey, health apps or devices, and GPS locator*. The independent variables in the models were age, educational status, occupational status, marital status, household income, mothers' health status, child health status, health care provider (HCP) status, children's age, number of children, use of internet on the mobile phone to access health information, mothers' motivation to share their data, and mothers' concerns about sharing health data. We generated odds ratios (ORs) with 95% Cis across levels of independent variables. *P* values <.05 were considered statistically significant at the 95% confidence level for the OR.

Results

Demographic Characteristics

Table 1 displays the demographic characteristics of non-Hispanic white mothers who responded to the survey questions. The survey results showed that the majority of the mothers were married (485/622, 78.0%) women aged between 18 and 49 years (444/622, 71.4%) who had an excellent-to-good health condition (560/622, 90.0%). Regarding the occupational status of the participants, a large number of the investigated mothers were employed or self-employed (452/622, 72.7%). More than half of the mothers had at least a 4-year college degree (347/622, 55.8%), with a yearly household income of over $75,000 (388/622, 62.4%). More than half of the mothers reported having 1 child (326/622, 52.4%), and the majority of their children or young adults were in excellent-to-good health status (594/622, 95.5%).

Mothers' motivation to share their data was split nearly equally between less motivated (302/622, 48.6%) and motivated (320/622, 51.4%). The mothers were motivated to share their data to contribute to science (326/622, 52.4%) and to benefit patient health (387/622, 62.2%)—these results are available upon request.

The majority of the respondents were concerned about the misuse of personal health information (507/622, 81.5%). In fact, the respondents were very concerned that their data would be stolen by unknown individuals or companies (360/622, 57.9%) or if their health data would be used without their consent and knowledge (340/622, 54.7%)—these results are available upon request.

Respondents were more willing to share their data with the researchers provided through Web-based surveys (509/622, 81.8%) and collected through their mobile phones (423/622, 68.0%) compared with their medical record data (224/622, 36.0%) and GPS locations (252/622, 40.5%).

Table 1. Frequency of mothers’ demographic characteristics (N=622).

Demographics	Values, n (%)
Age group (years)	
18-49	444 (71.4)
>50	178 (28.6)
Education	
Less than 4 years of a college degree	275 (44.2)
4 or more years of college	347 (55.8)
Occupational status	
Employed or self-employed	452 (72.7)
Other occupational status	170 (27.3)
Marital status	
Married	485 (78.0)
Other marital status	137 (22.0)
Household income (US$)	
≤74,999	234 (37.6)
≥75,000	388 (62.4)
Mothers’ health status	
Fair to poor	62 (10.0)
Excellent to good	560 (90.0)
Child health status	
Fair to poor	28 (4.5)
Excellent to good	594 (95.5)
HCP[a] status	
I don’t have	52 (8.4)
Have more than 1 HCP	190 (30.5)
Yes, just 1 HCP	380 (61.1)
Children’s age (years)	
≤14	381 (61.1)
≥15	241 (38.7)
Number of children	
One child	326 (52.4)
More than one child	296 (47.6)
Use of mobile phones	
Yes	553 (96.0)
No	21 (3.4)
Use of the internet to access health information[b]	
Yes	361 (58.0)
No	251 (40.4)
Mothers’ motivation for sharing their data	
Less motivated	302 (48.6)
Somewhat motivated	248 (39.9)
Very motivated	72 (11.6)

Demographics	Values, n (%)
Mothers' concerns about the misuse of personal health information	
Less concerned	115 (18.5)
Somewhat concerned	227 (36.5)
Very concerned	280 (45.0)
Utilization of patient portals	
Never used it	228 (36.7)
Used it more than once	394 (63.3)
Desire to keep personal health information anonymous	
Not at all	13 (2.1)
Somewhat	169 (27.2)
Extremely	440 (70.7)
Willingness to share anonymous data from medical records	
Yes	224 (36.0)
No	398 (64.0)
Willingness to share anonymous data provided through a Web-based survey	
Yes	509 (81.8)
No	113 (18.1)
Willingness to share anonymous data entered into or collected by health-related app or device	
Yes	423 (68.0)
No	199 (32.0)
Willingness to share GPS location anonymously from a mobile app	
Yes	252 (40.5)
No	370 (59.5)

[a] HCP: health care provider.

[b] 48 participants did not respond to this question.

Associations of Mothers' Desire to Keep Their Health Information Anonymous and Independent Variables

The chi-square test results showed that the HCP status (*P*=.02), the use of the internet to access health information (*P*=.03), mothers' motivation (*P*=.01), and concerns (*P*<.001) about sharing health data were all associated with mothers' desires to keep their data anonymous (Multimedia Appendix 1). After adjusting for the demographic characteristics of mothers using multivariate logistic regression, only mothers' concerns about sharing data were associated with the desire to keep health information anonymous (Table 2). Mothers who were very concerned and somewhat concerned were more than two times and four times more likely to keep their health information anonymous than less-concerned mothers (adjusted odds ratio [aOR] 4.77, 95% CI 2.85-7.96; aOR 2.50, 95% CI 1.52-4.1, respectively).

Table 2. Results of multivariate logistic regression regarding the association between mothers' desire to keep their health information anonymous and a set of predictors, including their demographic characteristics.

Effect	aOR[a] (95% CI)	*P* value[b]
Age (years)		**.86**
18-49	0.95 (0.55-1.64)	
>50	1.00 (Reference)[c]	
Educational level		**.17**
4 or more years of college	0.75 (0.5-1.13)	
Less than 4 years of a college degree	1.00 (Reference)[c]	
Occupational status		**.20**
Employed	1.34 (0.86-2.1)	
Unemployed	1.00 (Reference)[c]	
Marital status		**.35**
Married	1.27 (0.77-2.1)	
Unmarried	1.00 (Reference)[c]	
Income level (US $)		**.42**
≥75,000	1.19 (0.77-1.84)	
≤74,999	1.00 (Reference)[c]	
Health status		**.93**
Excellent to good	1.03 (0.52-2.03)	
Fair to poor	1.00 (Reference)[c]	
Child health status		**.36**
Excellent to good	0.61 (0.21-1.75)	
Fair to poor	1.00 (Reference)[c]	
HCP[d] status		**.50**
More than 1 HCP	0.82 (0.37-1.84)	
Just 1 HCP	0.69 (0.32-1.47)	
No HCP	1.00 (Reference)[c]	
Children's age (years)		**.99**
≤14	1.00 (0.59-1.7)	
≥15	1.00 (Reference)[c]	
Number of children		**.98**
More than one child	1.01 (0.66-1.54)	
One child	1.00 (Reference)[c]	
Use of mobile phones		**.14**
Yes	2.06 (0.8-5.33)	
No	1.00 (Reference)[c]	
Use of the internet to access health information		**.33**
Yes	0.35 (0.04-2.91)	
No	1.00 (Reference)[c]	
Mothers' motivation to share their data		**.11**
Somewhat motivated	0.66 (0.43-1)	

Effect	aOR[a] (95% CI)	*P* value[b]
Very motivated	0.66 (0.36-1.22)	
Less motivated	1.00 (Reference)[c]	
Mothers' concern about sharing health data		**<.001**
Somewhat concerned	2.50 (1.52-4.1)	
Very concerned	4.77 (2.85-7.96)	
Less concerned	1.00 (Reference)[c]	

[a] Adjusted odds ratios (aORs) of reporting desire to keep health information anonymous from a multivariable logistic regression model, conditional on mothers' motivation and concerns and all other characteristics

[b] *P* values from Type 3 analysis based on the Wald test.

[c] Reference group does not have CI.

[d] HCP: health care provider.

Associations of Mothers' Use of Patient Portals With Independent Variables

Bivariate analyses indicated that child health status (*P*=.03), status of HCP (*P*<.001), use of mobile phones (*P*<.001), use of the internet to access health information (*P*<.001), and mothers' motivation to share data (*P*<.001) were statistically associated with mothers' use of the patient portal (Multimedia Appendix 1). However, after adjusting for mothers' demographic characteristics using multivariate logistic regression models, only HCP status and mothers' motivation to share data had statistically significant associations with the utilization of the patient portal (Table 3). The likelihood of using patient portals increased four times for mothers who had 1 HCP (aOR 3.47, 95% CI 1.73-6.94) and more than one HCP (aOR 4.3, 95% CI 2.06-8.99). Similarly, very motivated mothers who were interested in sharing their health data used the patient portal twice as much as other investigated mothers (aOR 2.09, 95% CI 1.12-3.91).

Table 3. Results of multivariate logistic regression regarding the relationship between mothers' use of the patient portal and a set of predictors, including mothers' demographic characteristics.

Effect	aOR[a] (95% CI)	*P* value[b]
Age (years)		**.54**
18-49	0.86 (0.52-1.41)	
>50	1.00 (Reference)[c]	
Education level		**.92**
4 or more years of college	0.98 (0.67-1.44)	
Less than 4 years of a college degree	1.00 (Reference)[c]	
Occupational status		**.54**
Employed	0.87 (0.57-1.35)	
Unemployed	1.00 (Reference)[c]	
Marital status		**.91**
Married	0.97 (0.6-1.58)	
Unmarried	1.00 (Reference)[c]	
Income level (US $)		**.11**
≥75,000	1.40 (0.93-2.12)	
≤74,999	1.00 (Reference)[c]	
Health status		**.70**
Excellent to good	0.88 (0.46-1.68)	
Fair to poor	1.00 (Reference)[c]	
Child health status		**.13**
Excellent to good	0.43 (0.14-1.3)	
Fair to poor	1.00 (Reference)[c]	
HCP[d] status		**<.001**
More than one HCP	4.3 (2.06-8.99)	
Just one HCP	3.47 (1.73-6.94)	
No HCP	1.00 (Reference)[c]	
Children's age (years)		**.32**
≤14	1.29 (0.79-2.11)	
≥15	1.00 (Reference)[c]	
Number of children		**.52**
More than one child	1.14 (0.76-1.72)	
One child	1.00 (Reference)[c]	
Use of mobile phones		**.15**
Yes	2.01 (0.77-5.2)	
No	1.00 (Reference)[c]	
Use of the internet to access health information		**.26**
Yes	2.27 (0.55-9.32)	
No	1.00 (Reference)[c]	
Mothers' motivation to share their data		**.006**
Somewhat motivated	1.75 (1.18-2.59)	

Effect	aOR[a] (95% CI)	P value[b]
Very motivated	2.09 (1.12-3.91)	
Less motivated	1.00 (Reference)[c]	
Mothers' concern about sharing health data		**.26**
Somewhat concerned	1.43 (0.85-2.39)	
Very concerned	1.46 (0.88-2.42)	
Less concerned	1.00 (Reference)[c]	

[a] Adjusted odds ratios (aORs) of reporting use of patient portals from a multivariable logistic regression model, conditional on mothers' motivation and concerns and all other characteristics.

[b] *P* values from Type 3 analysis based on the Wald test.

[c] Reference group does not have CI.

[d] HCP: health care practitioner.

Mothers' Willingness to Share Their Data From Different Sources

Association of Mothers' Willingness to Share Data Through Electronic Medical Record Data and Independent Variables

The results of the bivariate analysis indicated that mothers' willingness to share their electronic medical record (EMR) data was significantly related to mothers' marital status (*P*=.01), household income (*P*=.04), child health status (*P*=.02), use of the internet to access health information (*P*=.02), motivation (*P*=.02), and privacy concerns (*P*=.02) about sharing health data (Multimedia Appendix 1).

When running the multivariate logistic regression analysis (Table 4), the variables that were statistically significantly associated with the willingness to share EMR data anonymously included mothers' motivation to share data, mothers' concern about sharing their health data, child health status, and HCP status. The results showed that mothers with a child in excellent-to-good health status (aOR 0.38, 95% CI 0.15-0.93) were less likely to share EMR data anonymously. Furthermore, mothers with one HCP (aOR 1.61, 95% CI 0.95-2.73) increased their likelihood of willingness to share their EMR data with researchers by 2.7 times. Additionally, very motivated mothers were nearly four times more willing to share their EMR data with researchers (aOR 3.64, 95% CI 2.00-6.63). Highly concerned mothers were 40% less likely to share EMR data with researchers, compared with mothers who were less concerned about sharing data.

Association of Mothers' Willingness to Share Data Provided in a Web-Based Survey With Independent Variables

The bivariate analysis revealed that the status of HCP (*P*<.001), use of mobile phones (*P*=.01), use of the internet to access health information (*P*<.001), and mothers' motivation to share data (*P*<.001) were statistically associated with mothers' willingness to share their data in Web-based surveys (Multimedia Appendix 1). However, after adjusting for mothers' characteristics in multivariate analysis, only the status of HCP (*P*<.001) and mothers' motivation to share data (*P*<.001) were statistically associated with their willingness to share data through a Web-based survey (Table 4). Mothers who had 1 (aOR 4.22, 95% CI 1.97-9.05) or more than one HCP (aOR 4.47, 95% CI 1.94-10.3) were four times as likely to share their health data in Web-based surveys with researchers.

Association of Mothers' Willingness to Share Data Entered in Health Apps or Collected From Devices and Independent Variables

The findings of this study indicated that the use of mobile phones (*P*<.001), use of the internet for health information (*P*<.001), mothers' motivation to share data (*P*<.001), and mothers' concerns about sharing data (*P*<.001) were statistically associated with their willingness to share mobile app data in bivariate analyses (Multimedia Appendix 1). The results from multivariate logistic regression analyses showed that younger mothers (aOR 1.93, 95% CI 1.12-3.32) were nearly two times more willing to share their app or device data with researchers than older mothers (Table 4). Mothers who had 1 (aOR 3.64, 95% CI 1.73-7.65) and more than one HCP (aOR 3.03, 95% CI 1.38-6.67) were three times more likely to share the health data of their apps or devices, compared with the reference group. Mothers who used smartphones (aOR 5.47, 95% CI 1.76-17) were five times more willing to share their data than the reference group.

Association of Mothers' Willingness to Share GPS Location Data and Independent Variables

The independent variables of children's age (*P*=.01), use of mobile phones (*P*=.03), access to health information (*P*<.001), mothers' motivation (*P*<.001), and concerns (*P*<.001) were statistically associated with mothers' willingness to share their GPS location in bivariate analyses (Multimedia Appendix 1).

Table 4. Results of multivariate logistic regression regarding mothers' willingness to share different types of data with researchers and a set of predictors, including mothers' demographic characteristics.

Effect	Willingness to share anonymous data from							
	Electronic medical data		Web-based surveys		Health app and device		GPS	
	aOR[a] (95% CI)	*P* value[b]	aOR (95% CI)	*P* value[b]	aOR[a] (95% CI)	*P* value[b]	aOR[a] (95% CI)	*P* value[b]
Age (years)		**.52**		**.80**		**.02**		**.84**
18-49	0.84 (0.49-1.43)		1.09 (0.57-2.07)		1.93 (1.12-3.32)		1.05 (0.63-1.76)	
>50	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	
Education level		**.53**		**.14**		**.56**		**.84**
4 or more years of college	1.14 (0.76-1.7)		0.68 (0.41-1.14)		1.13 (0.74-1.73)		1.04 (0.70-1.54)	
Less than 4 years of a college degree	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	
Occupational status		**.53**		**.18**		**.84**		**.24**
Employed	1.15 (0.74-1.81)		0.66 (0.36-1.21)		0.95 (0.59-1.53)		1.3 (0.84-2.02)	
Unemployed	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	
Marital status		**.14**		**.66**		**.79**		**.82**
Married	0.69 (0.42-1.12)		0.86 (0.44-1.68)		0.93 (0.53-1.63)		0.95 (0.59-1.53)	
Unmarried	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	
Income level (US $)		**.16**		**.82**		**.32**		**.06**
≥75,000	0.73 (0.48-1.13)		1.07 (0.61-1.86)		0.79 (0.49-1.27)		0.67 (0.44-1.01)	
≤74,999	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	
Health status		**.23**		**.27**		**.59**		**.66**
Excellent to good	0.67 (0.36-1.28)		1.57 (0.71-3.49)		1.21 (0.61-2.42)		0.87 (0.46-1.62)	
Fair to poor	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	
Child health status		**.03**		**.51**				**.28**
Excellent to good	0.38 (0.15-0.93)		1.51 (0.45-5.12)		1.31 (0.5-3.48)	.58	0.61 (0.25-1.5)	
Fair to poor	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	.59	1.00 (Reference)[c]	
HCP[d] status		**.06**		**<.001**		**.003**		**.18**
More than one HCP	2.23 (0.92-5.43)		4.47 (1.94-10.3)		3.03 (1.38-6.67)		2.11 (0.93-4.8)	
Just one HCP	2.69 (1.15-6.27)		4.22 (1.97-9.05)		3.64 (1.73-7.65)		2.04 (0.93-4.5)	
No HCP	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	
Children's age (years)		**.07**		**.37**		**.94**		**.07**
≤14	1.61 (0.95-2.73)		1.35 (0.7-2.6)		1.02 (0.58-1.79)		1.6 (0.96-2.66)	
≥15	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	
Number of children		**.41**		**.78**		**.84**		**.06**
More than one child	0.84 (0.55-1.28)		0.92 (0.53-1.61)		0.95 (0.6-1.52)		0.68 (0.44-1.02)	

Effect	Willingness to share anonymous data from							
	Electronic medical data		Web-based surveys		Health app and device		GPS	
	aOR[a] (95% CI)	*P* value[b]	aOR (95% CI)	*P* value[b]	aOR[a] (95% CI)	*P* value[b]	aOR[a] (95% CI)	*P* value[b]
One child	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	
Use of mobile phones		**.90**		**.06**		**<.001**		**.22**
Yes	1.07 (0.36-3.19)		2.85 (0.97-8.41)		5.47 (1.76-17)		2.01 (0.63-6.94)	
No	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	
Use of the internet to access health information		**.61**		**.70**		**.69**		**.44**
Yes	1.58 (0.27-9.27)		1.37 (0.28-6.7)		0.73 (0.16-3.4)		1.93 (0.35-10.69)	
No	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	
Mothers' motivation to share their data		**<.001**		**<.001**		**<.001**		**<.001**
Somewhat motivated	2.42 (1.6-3.67)		5.94 (3.15-11.2)		3.37 (2.16-5.26)		3.1 (2.08-4.60)	
Very motivated	3.64 (2.00-6.63)		2.87 (1.18-6.94)		5.30 (2.32-12.1)		5.15 (2.83-9.38)	
Less motivated	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	
Mothers' concern about sharing health data		**.008**		**.31**		**.09**		**<.001**
Somewhat concerned	0.59 (0.35-1.00)		0.75 (0.34-1.68)		0.88 (0.47-1.68)		0.50 (0.27 0.77)	
Very concerned	0.43 (0.25-0.73)		0.57 (0.26-1.25)		0.58 (0.31-1.08)		0.40 (0.22-0.60)	
Less concerned	1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]		1.00 (Reference)[c]	

[a] Adjusted odds ratios (aORs) of reporting willingness to share different type of data from a multivariable logistic regression model, conditional on mothers' motivation and concerns and all other characteristics.

[b] *P* values from Type 3 analysis based on the Wald test.

[c] Reference group does not have CI.

[d] HCP: health care provider.

When adjusting for mothers' characteristics through multivariate logistic regression, only mothers' motivation to share data (*P*<.001) and mothers' concerns with sharing data (*P*<.001) were statistically associated with their willingness to share their GPS location (Table 4). Furthermore, very motivated mothers (aOR 5.1, 95% CI 2.83-9.38) were five times more likely to be willing to share their GPS location data. Similarly, somewhat motivated mothers (aOR 3.1, 95% CI 2.08-4.60) were three times more willing to share their GPS location data than the reference group. Conversely, very concerned mothers (aOR 0.4, 95% CI 0.22-0.60) were 40% less willing to share their data, compared with less concerned ones.

Discussion

This study explored mothers' motivation to share health data, concerns with potential misuse of personal health information, and willingness to share different types of data with researchers, their utilization of patient portals, and their desire to keep their health information anonymous.

Motivation to Share Data and Concern With Data Use

Our study results revealed that about half of the mothers were less motivated to share their data with researchers. Our results contradict the findings of a previous study that found that more than 78% of the surveyed respondents were more willing to share their data with researchers [25]. However, when we investigated the reasons driving these mothers to share their health data, we found that they were motivated to contribute to science and benefit other patients. On the other hand, we found that 55% of mothers were concerned with the misuse of their personal health information. A major concern with the misuse of personal health data has been reported among 68% of healthy volunteers in survey studies [28]. Further analysis showed that their privacy concerns were related to data misuse, especially the risk that their data would be stolen or used without their consent. In other words, their concern impacted their perceived risk of privacy for disclosing their data, which is consistent with previous studies [18].

Desire to Keep Health Information Anonymous and Use of Patient Portal

Our findings on the respondents’ desire to keep their health information anonymous have also been reported elsewhere [29]. Moreover, we found that mothers’ desire to keep their personal health information anonymous was dependent on their perceived concerns. These results were in line with previous studies on the benefits and concerns of data sharing [25].

We found that a relatively high proportion of patient portal use (63%) among women in the general population may seem unusual. Although health portal use by patients is becoming more prevalent, a recent study estimated that only 32% of outpatients of a Dutch academic health center used a patient portal [30]. Another study reported a 58% registration rate to the patient portal among older adult patients linked to an academic medical center in the United States [31]. However, more recent studies have shown a much higher percentage of portal users among adult patients (82%) in the United States [32]. In addition, another study identified that 34% of Americans have been offered access to patient health information through an HCP, but only 28% accessed this information [33].

Willingness to Share Anonymous Data

Our study found that the majority of the mothers were not willing to share anonymous data from medical records and their GPS location using their mobile app (or device). However, these mothers were willing to share anonymous data through a Web-based survey. Our review of the literature cannot corroborate these findings as most of the studies on individuals’ willingness to share their health data focused on health information exchanges [11,12,14-16,18-21]. These studies found that data security and privacy preferences shape consumers’ attitudes toward health information exchanges. Moreover, consumers perceived health information exchanges to confer benefits, such as better coordination of care [15] and improved health care quality [11]. It has been reported that a person’s willingness to share health data is directly associated with the subject suffering from progressive or chronic illness [34]. However, our respondents’ or their children’s health status were not associated with their willingness to share data. Other studies on sharing patient health data reported conflicting results. One strand of studies has shown that individuals are willing to share their data to benefit health outcomes [35], whereas another strand found that anonymity, research use, engagement with a trusted entity, transparency to access medical records, and incentives affected individuals’ willingness to share their data [22,29]. The findings of this study confirmed the latter strand, indicating that the level of mothers’ concerns played a more significant role than their motivations in sharing their medical records. In fact, mothers were concerned that their data would be misused or stolen. With regard to participants sharing GPS location, studies have reported low willingness to share location data [36,37]. The results from these two studies conform with our findings, and according to a study report, sharing the GPS location can jeopardize the privacy and personal information of patients [38].

Strengths and Limitations

To our knowledge, this is the first study that employed the opt-in panel to assess non-Hispanic white mothers’ attitudes and perceptions toward data sharing [4]. As the opt-in panel is a self-selected sample of women, matched to the background of mothers of children with autism, our results about health data sharing behavior can only be generalized to non-Hispanic white mothers in the same age, education, and employment status group of our study sample. Owing to this study design, our findings are not representative of nonwhite single women in the United States or other populations in the United States. Moreover, we should interpret our results with caution when compared with similar studies due to the nature of our survey questions. In particular, previous studies have framed patient health data entirely from the health information exchange perspective, and more specifically, those studies investigated data sharing with care providers but not with researchers. Finally, the obtained results of some of our statistical analyses were too small; therefore, statistical testing can be unstable.

Future Research Implications

In an era dominated by mobile apps and wearable devices, researchers should focus on the value of the privacy calculus in the context of data sharing for research [26,39,40]. First, this interest is rising with the spread of ubiquitous computing and unlimited options for collecting, processing, distributing, and using data, which can overwhelm participants’ interest in sharing their data. On the other hand, the future of health care discoveries rests upon the amount of data collected from patients. In this regard, many participants may not know how their data have been used and accessed. Therefore, this lack of clear communication among members of the research community and the general population on how data are being collected and used may raise ethical issues related to data sharing [23].

Second, to facilitate and improve participation in citizen research, which requires recruiting a large number of individuals to participate in a health research study, a priori market segmentation studies should be implemented to assess consumers’ data-sharing behavior. Such methods are more rigorous than extrapolating the findings from the general population. Consumers’ data-sharing behavior is warranted in part because of the digital divide that is due to the difference in socioeconomic status exhibited within the general population [11,33,41,42]. Previous studies have suggested that increasing consumers’ trust in information communication technology and data sharing can lead to higher participation in data-sharing research [18]. The findings of this study suggest that researchers studying data-sharing behavior should have a better understanding of their targeted group so that they can identify strategies to increase their participation. Furthermore, our findings suggest the need to engage patients in addressing the underlying reasons for their concerns. Finally, our findings are aligned with previous research that recommended assessing consumers’ data-sharing behavior. This assessment can provide guidelines for Web and apps development that can provide consumers with better access and control over their data, which can subsequently increase consumers’ trust [18,22].

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Conflicts of Interest

None declared.

Multimedia Appendix 1
Supplemental results (bivariate analysis).
[DOCX File , 43 KB - jopm_v12i2e14062_app1.docx]

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Abbreviations

aOR: adjusted odds ratio
ASD: autism spectrum disorder
eHealth: electronic health
EMR: electronic medical record
HCP: health care provider
IAN: Interactive Autism Network
OR: odds ratio

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Original Paper

Methicillin-Resistant Staphylococcus aureus Eradication and Decolonization in Children Study (Part 1): Development of a Decolonization Toolkit With Patient and Parent Advisors

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Abstract

Background: Community-acquired methicillin-resistant *Staphylococcus aureus* (MRSA) skin and soft tissue infections affect many healthy children. A significant number of these children are hospitalized and require surgical incision and drainage (I&D). Once sent home, these children and families are asked to complete burdensome home decolonization and hygiene procedures in an effort to prevent the high rate of recurrent infections.

Objective: This component of the Methicillin-resistant *Staphylococcus aureus* Eradication and Decolonization in Children (MEDiC) study aimed to develop a toolkit to assist MEDiC study participants in completing MRSA decolonization and hygiene procedures at home (the MEDiC kit).

Methods: In all, 5 adolescents (aged 10-18 years) who had undergone an I&D procedure for a skin infection and 11 parents of children who had undergone an I&D procedure for a skin infection were engaged in a 4-hour group workshop using a human-centered design approach. The topics covered in this workshop and analyzed for this paper were (1) attitudes about MRSA decolonization procedures and (2) barriers to the implementation of MRSA decolonization and hygiene procedures. The team analyzed the audio and artifacts created during the workshop and synthesized their findings to inform the creation of the MEDiC kit.

Results: The workshop activities uncovered barriers to successful completion of the decolonization and hygiene procedures: lack of step-by-step instruction, lack of proper tools in the home, concerns about adverse events, lack of control over some aspects of the hygiene procedures, and general difficulty coordinating all the procedures. Many of these could be addressed as part of the MEDiC kit. In addition, the workshop revealed that effective communication about decolonization would have to address concerns about the effects of bleach, provide detailed information, give reasons for the specific decolonization and hygiene protocol steps, and include step-by-step instructions (preferably through video).

Conclusions: Through direct engagement with patients and families, we were able to better understand how to support families in implementing MRSA decolonization and hygiene protocols. In addition, we were able to better understand how to communicate about MRSA decolonization and hygiene protocols. With this knowledge, we created a robust toolkit that uses patient-driven language and visuals to help support patients and families through the implementation of these protocols.

Trial Registration: ClinicalTrials.gov NCT02127658; https://clinicaltrials.gov/ct2/show/NCT02127658

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KEYWORDS

Staphylococcus aureus; MRSA; abscess; decolonization; human-centered design; communication design

Introduction

Background

The past two decades have seen a dramatic increase in community-acquired skin and soft tissue infections (SSTIs), such as cellulitis, boils, myositis, and abscesses, caused by the antibiotic-resistant bacteria known as methicillin-resistant *Staphylococcus aureus* (MRSA) [1-8]. The shift from hospital-acquired infections to community-acquired infections has resulted in many healthy children being affected [2,4]. Recent estimates indicate that the incidence of hospitalizations in the United States caused by MRSA SSTIs is more than 45 per 100,000 children, with many children requiring surgical procedures such as incision and drainage (I&D) [6,9,10]. The rate of recurrent infection can be as high as 72% [11-16].

As frequent recurrence of MRSA SSTIs is believed to increase suffering, health care utilization, and cost, strategies to decrease the rate of recurrence are necessary. MRSA colonization (presence of bacteria on the skin and in the nose) has been demonstrated to be a risk factor for SSTIs and their recurrence, and there has been a high prevalence of MRSA colonization among patients presenting to emergency departments in the United States [17,18]. Decolonization protocols using topical mupirocin ointment in the nose to eliminate nasal carriage and chlorhexidine or bleach baths to eliminate skin carriage [19-21] are often recommended by the Infectious Disease Society of America as strategies to eradicate the bacteria and decrease the recurrence of hospital-acquired MRSA infections [11]. The prevention of community-acquired MRSA has been studied to a much lesser degree and the success rates of these protocols are mixed [11,15]. Additionally, few studies have addressed the burden of these decolonization protocols that often consist of regular bleach baths or chlorhexidine body washes and daily nasal antibiotics, on patients and their families. One study that aimed to better understand the feasibility of decolonization protocols found that only 38% of participants reported having completed both components of a 5-day protocol involving twice daily mupirocin nasal swabs and once daily diluted bleach baths. Some barriers reported by participants were side effects such as skin irritation, aversion to the smell of bleach, and being too busy [22]. This study sheds some light on barriers that may exist in MRSA decolonization, but is in the context of only a very short 5-day protocol and does not shed light on how these barriers might be addressed.

Objectives

With the aid of patients and families with lived experiences with MRSA SSTIs, we sought to design the Methicillin-resistant *Staphylococcus aureus* Eradication and Decolonization in Children (MEDiC) comparative effectiveness trial assessing 2 interventions over the course of 12 months: (1) abscess surgery and hygiene education compared with (2) abscess surgery and hygiene education followed by decolonization [23]. Knowing that decolonization procedures can be burdensome, we engaged patients and their families (referred to as advisors herein) in a human-centered design (HCD) workshop to understand (1) their attitudes about MRSA decolonization procedures and (2) uncover potential barriers to the implementation of MRSA decolonization and hygiene procedures. This paper will discuss the workshop activities, results, and how our findings informed the creation of a *toolkit* to assist participants in the MEDiC intervention study with decolonization and hygiene procedures. A companion paper discusses a separate objective of this project, which was to engage patients and their families to better understand what outcomes were important to them when it came to MRSA decolonization and to select measures to capture these as part of the MEDiC study [24].

Methods

Overview

This patient engagement project was the first step in designing an MRSA decolonization toolkit to prepare for a randomized comparative effectiveness trial (MEDiC-NCT02127658), as described briefly earlier. This study was approved by the Indiana University School of Medicine Institutional Review Board. Participants were eligible if they were a patient (aged 9-18 years) who had undergone an I&D procedure at Riley Hospital for Children or a parent of a patient over 3 months of age who had undergone an I&D procedure at Riley Hospital for Children. Written informed consent was obtained from all participants over the age of 13 years. Participants aged 9 to 13 years provided assent. All participants received US $20 per hour for the 4-hour audio-recorded workshop.

Human-Centered Design

The Indiana Clinical and Translational Sciences Institute’s patient engagement core, known as Research Jam (RJ), uses an HCD approach to co-design better research study experiences with study stakeholders. HCD developed out of the ergonomics and computer science [25] fields and has expanded widely to be used in the creation of a vast array of products, services, and experiences across many different fields. HCD is a qualitative approach to understanding human needs, designing solutions that address these needs, and doing so hand in hand with stakeholders throughout the process. Stakeholders are considered experts in the problem area and are engaged as advisors and co-designers in the HCD process. The involvement of stakeholders is generative (allowing them to create the possibilities) rather than solely evaluative (allowing them to respond to predefined possibilities). RJ collaborated with the principal investigator (PM) to explore potential barriers the kit would need to overcome as well as desired messages (the framing of the information) and media (the format through which the information is delivered) and apply this knowledge to create the MEDiC kit. The workshop, facilitated by HCD experts DL and CM, and using the principles of HCD, comprised the following 3 methods.

Method I: Media Warm-Up

To begin, the participants (who will be referred to as advisors) were each asked to share their name and two ways they like to get information. The answers to the latter were written on a flip chart. This accomplished 2 goals: (1) to allow the advisors to get acquainted with one another and comfortable speaking aloud to the group and (2) to understand the preferred method of obtaining information, setting a foundation for further discussion.

Method II: Task Analysis of Methicillin-Resistant Staphylococcus Aureus Decolonization and Hygiene

The team used a simplified version of hierarchical task analysis, which examines the tasks needed to achieve a goal, ultimately informing strategies that enable effective use of systems or products. Task analysis is often used to iterate operations within a system, frequently resulting in changes to the tasks themselves [26]. In our case, the tasks to be analyzed are an accepted protocol, and so our focus was not on changing the tasks but changing the artifacts designed to support the tasks and to support behavioral changes. Rather than diving deeply into each task as is possible with task analysis, we chose to focus on breadth across the varied tasks in hopes to better understand the challenges posed by the broader personal, family, and environmental contexts in which these tasks might be undertaken. In this way, we hoped to create support artifacts that respond to these challenges and help families adhere more successfully to the study protocol. We separated the MEDiC study intervention goal of MRSA decolonization into 12 proposed tasks:

1. Uniforms and practice jerseys should be washed after each game or practice. Other sports equipment should be cleaned weekly.
2. Do not share towels, washcloths, clothing, toothbrushes, or razors with family or friends.
3. Wash all towels, washcloths, sleepwear, underwear, and linens in detergent and hot water once weekly and dry hot in a dryer.
4. Take daily showers or baths with soap.
5. Shower before and after all sports practices and competitions and wipe down all equipment before and after use.
6. Clean hands with soap and water (or hand sanitizer) when hands are dirty and after bathroom breaks and diaper changes.
7. Apply 2% mupirocin ointment to nostrils using a cotton swab twice a day for 10 days.
8. Twice weekly for 6 weeks, take a bath for 15 min in diluted bleach water.
9. Diluted bleach water should be made with 1 cup of 6% sodium hypochlorite bleach for a 50-gallon bathtub of water or 1 tsp per gallon of water.
10. Keep all wounds, including cuts and scrapes, clean and covered until healed.
11. Avoid other people’s dirty bandages or uncovered wounds.
12. Throw away all lotions in jars.

Advisors were asked to review the tasks and identify pain points—potential moments of difficulty—and brainstorm ways to overcome these pain points. Each task was displayed on a flipchart. Advisors were asked *What might make it hard to do this?* and responded by writing on individual post-it notes as many responses as they could think of. For the first round of responses, the advisors were asked to individually write their answers on post-it notes and add these post-its to the tasks to which they referred. For the second round of brainstorming, facilitators (CM and DL) read the answers aloud to the group and asked follow-up questions about the content of the notes to clarify meaning and encourage discussion to uncover additional pain points. Once the advisors had no other pain points to contribute, the facilitator asked advisors to brainstorm ways to overcome the major pain points, and these were added to the flipchart pages. The facilitator referred advisors to the flipchart from the media warm-up containing ways they liked to get information to help prompt additional ideas.

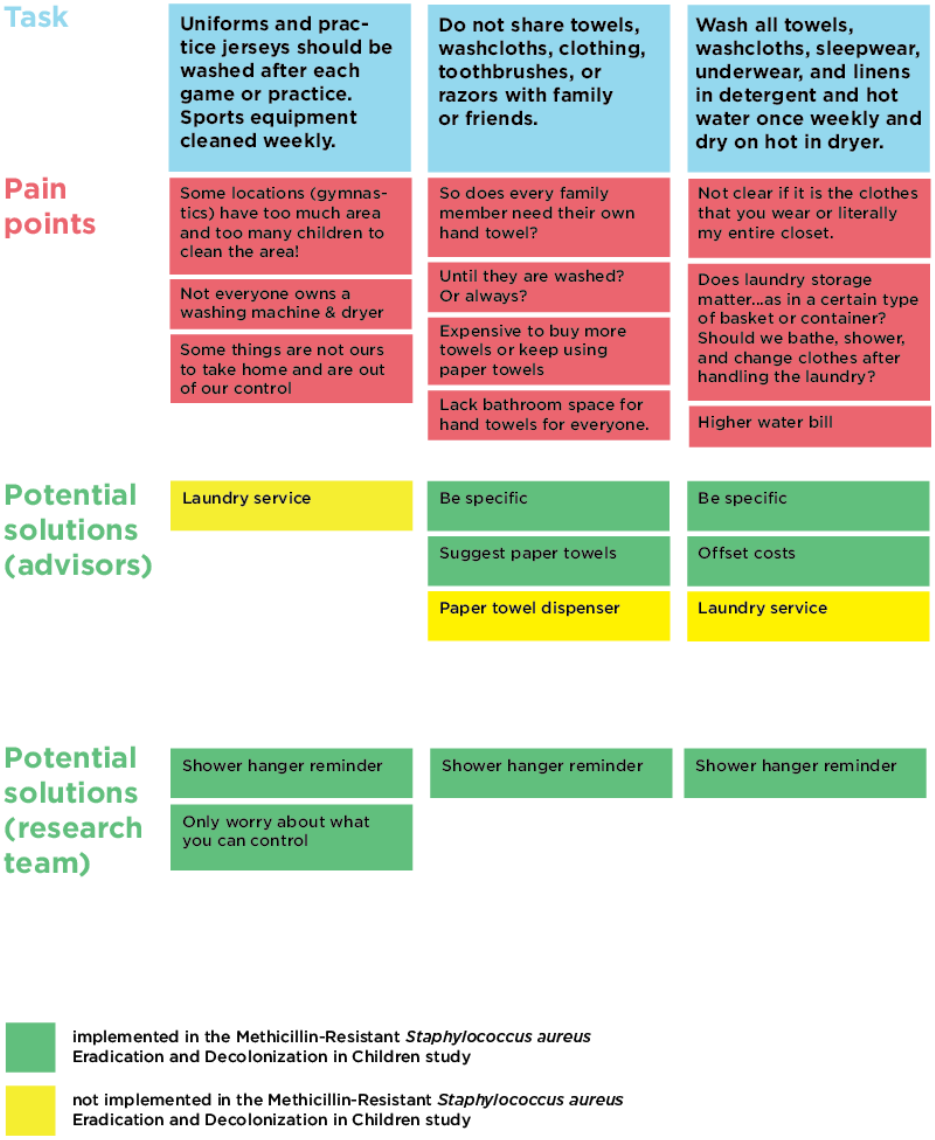
Method III: Five Senses Maps

A Five Senses map is used to display sensory information that a particular subject evokes. Typically, the subject is a brand, and the sensory information is aspirational—senses the brand should evoke with its customers through all aspects of its interactions with them, called sensory marketing [27]. In the context of this project, the Five Senses map visualization was used as a tool to structure our questioning, keep responses visible to the advisors, and allow the advisors to collaboratively build early maps during the workshop that would serve as prototypes for later maps. The goal of this 3-part activity was to understand the advisors’ sensory experience of bleach, both positive and negative. Understanding the perceptual barriers to accepting and following a bleach bath protocol would allow the team to craft messaging or make adjustments to the protocol to help overcome these perceptions. The first part of the activity asked the advisors—1 sense at a time—to talk about the smells, sounds, tastes, sights, and tactile feelings bleach brought to mind. These were written by a facilitator CM on a flip chart in the form of a Five Senses map. The second part of the activity was to look at the senses captured on the Five Senses map for bleach and suggest opposite senses to create a new Five Senses map that represents the exact opposite of bleach. This helped the team further interpret and define what the advisors meant by the senses they associated with bleach. The last part of this activity asked the advisors to think about how they might change bleach to make it more like the opposite Five Senses map. These answers were captured on a flipchart.

Analysis

To prepare for analysis, the audio related to the Five Senses mapping activity was reviewed by CM and DL, and information not captured on the Five Senses maps was added to the maps. The task analysis responses were transcribed by CM into individual snippets of information. Audio related to the task analysis activity was also reviewed, and content missing from the written responses was transcribed by CM onto individual snippets of information. The data, information, knowledge, wisdom (DIKW) framework, popularized by Ackoff and expanded upon by Kolko, guided the analysis process for the workshop data. DIKW is a framework that organizes the evolution of findings from data (discrete symbols) to wisdom (development of increased value through the application of knowledge) [28,29]. Team members CM and DL were guided by Kolko's synthesis methods for jumping the gaps between each stage of DIKW [29]. Through affinity clustering, the snippets of data were examined to find similarity in meaning and then grouped into clusters to begin to organize the data to create meaning. These meaningful data are what Kolko defines as information. The information clusters were given names to capture the meaning represented by the data they contained. Next, CM and DL engaged in visual modeling, which Kolko suggests for moving from information to knowledge (principles, theories, or stories) and involves organizing the clusters from the previous step into visual structures that present hypotheses about their relationships with each other. These visual models were (1) a task analysis grid (a model of tasks, pain points, and potential solutions from the advisors) and (2) a final Five Senses map that captured the positive associations of bleach while shifting the negative associations. The team (CM and DL) then reviewed the affinity diagrams and visual models and identified insights or key patterns that, when combined with new knowledge developed through the analysis process, had implications for the design product (the MEDiC kit). On the basis of these insights and existing expertise and experience, the team (CM, DL, and PM) identified additional potential solutions to add to the task analysis grid (Figure 1) and created the design of the final MEDiC kit. This application of knowledge is defined by Kolko as wisdom.

Figure 1. Task analysis grid 1.



Task	Uniforms and practice jerseys should be washed after each game or practice. Sports equipment cleaned weekly.	Do not share towels, washcloths, clothing, toothbrushes, or razors with family or friends.	Wash all towels, washcloths, sleepwear, underwear, and linens in detergent and hot water once weekly and dry on hot in dryer.
Pain points	Some locations (gymnastics) have too much area and too many children to clean the area! Not everyone owns a washing machine & dryer Some things are not ours to take home and are out of our control	So does every family member need their own hand towel? Until they are washed? Or always? Expensive to buy more towels or keep using paper towels Lack bathroom space for hand towels for everyone.	Not clear if it is the clothes that you wear or literally my entire closet. Does laundry storage matter...as in a certain type of basket or container? Should we bathe, shower, and change clothes after handling the laundry? Higher water bill
Potential solutions (advisors)	🟨 Laundry service	🟩 Be specific 🟩 Suggest paper towels 🟨 Paper towel dispenser	🟩 Be specific 🟩 Offset costs 🟨 Laundry service
Potential solutions (research team)	🟩 Shower hanger reminder 🟩 Only worry about what you can control	🟩 Shower hanger reminder	🟩 Shower hanger reminder

🟩 implemented in the Methicillin-Resistant *Staphylococcus aureus* Eradication and Decolonization in Children study

🟨 not implemented in the Methicillin-Resistant *Staphylococcus aureus* Eradication and Decolonization in Children study

Results

Workshop Participation

We engaged a total of 16 workshop advisors, including 5 adolescents (3 females aged 10, 14, and 18 years; 2 males aged 14 and 17 years), all of whom had undergone an I&D procedure in addition to 11 total parents (10 females and 1 male); 4 of these were parents of the adolescents in attendance, and the other 7 were parents of younger children (aged 15 months to 5 years) who had also undergone an I&D procedure. Parents and their adolescent children and parents of younger children were split into separate small groups during the task analysis portion of the workshop. Adolescents remained in the room with their parents during the workshop.

Key Pain Points

Using the methodology described above, the following barriers, or *pain points*, were identified by the advisors in response to the 12 tasks proposed to help prevent MRSA recurrence. Figure 1 shows an example of the task analysis grid, which organizes each of the tasks in the decolonization and hygiene protocols and their associated pain points (as identified by the advisors). Potential pain point solutions offered by the advisors are also captured on the grid. The solutions in green were implemented as part of the kit, whereas those in yellow were deemed to be not feasible or not within the scope of this project (see Multimedia Appendices 1-3 for the remainder of the task analysis grid). This section discusses the key barriers we heard from the advisors as well as what we did to address each in the kit.

Current Decolonization Instructions Are Not User Friendly and Do Not Answer Advisors’ Questions

What We Heard

Some of the advisors were already using a bleach bath protocol recommended by a physician. They were given the majority of the instructions verbally, although some advisors had been given brief written instructions. The instructions were not consistent across the advisors. A few of the families reported that they had watched videos on YouTube that had been created by other parents to show the process step by step. They found these helpful and more user-friendly than instructions they had been given by their physicians, but they were unsure whether or not they could trust the information presented because it was not from a physician. They suggested a video of families going through a decolonization protocol step by step created in partnership with a doctor or another trusted source. During the warm-up activity, advisors discussed their preferred methods of obtaining new information. Video was the most popular method. Learning by doing (hands-on) was the second most popular. Other methods that were mentioned included being trained, getting notifications, reading instructions, drawing out a plan, getting text messages, and looking into all the options before choosing one for implementation.

For some parts of the protocol, the draft instructions presented to the advisors left too much to interpretation. For example, the advisors questioned whether or not they needed to wash all the clothing and linens they owned once a week or just those they had used. Additionally, they wondered if laundry should be stored and handled in a particular way and if they should shower after handling dirty laundry. Other similar questions were as follows:

1. Can 1 cotton swab be used for applying the ointment to the nostrils (1 side per nostril) or should 2 different cotton swabs be used?
2. Does everyone in the family need their own hand towels?
3. Can towels be shared after they are washed or does 1 towel have to always belong to the same person forever?
4. Can you never ever use jars of lotion, or is it just that we need to start fresh and spoon it out from now on?

The advisors wanted to understand what the instructions in the protocol were accomplishing. Discarding lotion in jars was one part of the protocol that inspired many comments. For some of the advisors, this would mean throwing a lot of money in the trash, so they wanted a clear explanation of why this was necessary. Once the reason was explained, the advisors understood, but without a clear reason, this just seemed wasteful.

What We Did

Through collaboration between content expert PM and communication designers DL and CM, the team developed detailed, user-friendly instructional materials (Multimedia Appendix 4) for hygiene and decolonization that responded to the informational needs and questions expressed by the advisors.

Because the advisors preferred visual methods of getting new information, the team created visuals to accompany the written instructions. In addition, the team created a step by step instructional video featuring a medical professional (AC; Multimedia Appendix 5).

The final kit includes much of the information advisors asked for during the task analysis discussion to help them better understand and adhere to the procedures. For some of the procedures, this included being more explicit (eg, specifying that only used clothing and linens ought to be washed once a week). This also included giving reasons for adhering to procedures with less obvious justification (eg, that lotions in jars are easily contaminated and that is why they ought to be avoided).

Families May Not Have the Proper Tools for Implementing Decolonization and Hygiene Procedures

What We Heard

Bleach packaging makes it difficult for families to implement decolonization protocols as recommended. Bleach comes in heavy bottles with large openings for pouring. This makes it very difficult to pour a small amount of bleach into a small measuring cup (assuming you have one on hand) to obtain the proper measure of bleach. To overcome this, the advisors suggested having a pour spout that slows down the flow of bleach, having a pump that dispenses a consistent amount of bleach each time, or using bleach tablets that you can drop into the bath. Families also may not know the volume of their bathtub and may not have an empty gallon jug on hand to measure it, which might force them to guess and have an unknown and inaccurate bleach-to-water ratio.

What We Did

To ensure that the MEDiC study participants had the right equipment to complete the study procedures, they were provided with a bucket, a large measuring cup, a small measuring cup to use for infant baths, bleach, wax crayon, mupirocin, and cotton swabs in addition to the printed instructional materials in the MEDiC kit. *Splash-less* bleach was chosen because it has a thicker consistency and can be poured more easily. A packing slip (Multimedia Appendix 6) included a visual list of all of the items study participants were intended to receive.

Some Tasks Were Out of the Control of Families

What We Heard

The advisors discussed the difficulty of adhering to some parts of the protocol that may not be within their control. This is particularly true of those parts of the protocol that involve gyms or sports equipment. In some sports, the area is too large to reasonably expect a family to wipe down (eg, gymnastics mats and floors). In other cases, sports equipment might not belong solely to the athlete and might be managed by another person, making it impossible for families to clean or launder them. In other cases—such as at a gym—the correct supplies may not be available for cleaning off equipment. In addition, children are often under the care of school staff during much of the day, making it difficult for caregivers to ensure that they are washing their hands properly, not removing bandages, not sharing objects with other children, etc. Additionally, because small children need help with hygiene practices such as applying bandages, it may be difficult for a caregiver to avoid a child's wounds. In other cases, children may be under the care of another parent in a different household, which might necessitate an extra set of all of the supplies and instructions and buy-in from the other parent. Children may also be under the care of a babysitter in their own home who would need to be properly trained to undergo the various protocol pieces.

What We Did

Parents were very concerned about their lack of ability to control some portions of the protocol. It can be challenging to perfectly adhere to procedures that require such careful control inside and outside of the home. The team included content to reassure parents that they did not need to have perfect control, but that they should strive to do their best (eg, to take precautions when helping small children with MRSA with their wounds). For the MEDiC study, the team wanted to ensure participants were adhering to the protocol without causing them unnecessary anxiety and that they were encouraged to be honest when tracking for the study. In addition, materials such as the instructional video could function as a tool for parents to disseminate knowledge to other caregivers such as grandparents and daycare providers as needed.

Decolonization and Hygiene Tasks Are Burdensome

What We Heard

The hygiene protocol requires keeping a lot of information, events, and supplies organized, which the advisors thought might be difficult for some families. Remembering all of the protocol requirements is difficult, and keeping track of which family member has done which part of the protocol on which day makes this all the more complicated. In addition, keeping track of which hand towel, bath towel, washcloth, toothbrush, etc, is whose can be very difficult in a large household with only one bathroom.

What We Did

The team created materials to function as reminders for MEDiC study participants. In all, 2 study task tracking booklets were created; one for the hygiene plus decolonization arm of the study and one for the hygiene-only arm (Multimedia Appendices 7 and 8). In addition, a hygiene shower hanger (Multimedia Appendix 9) was created to keep important hygiene tasks visible for all participants. In addition, the team included suggestions that might ease the burden of some of the procedures (eg, suggesting paper towels instead of individual bathroom hand towels for each family member).

Some of the Advisors Had Incorrect Assumptions About Methicillin-Resistant Staphylococcus Aureus

What We Heard

Throughout the workshop, the advisors shared information that they had heard about MRSA. Some advisors thought it was transmitted from parent to child through DNA or blood. Others thought it was a virus. Before the workshop, 2 of the mothers had thought it was only caused by diapers, and once their children were out of diapers, the MRSA would be gone. One mother suspected that her child contracted MRSA from being on the beach in Florida. These responses highlight a lack of basic education about MRSA, even for families experiencing it firsthand.

What We Did

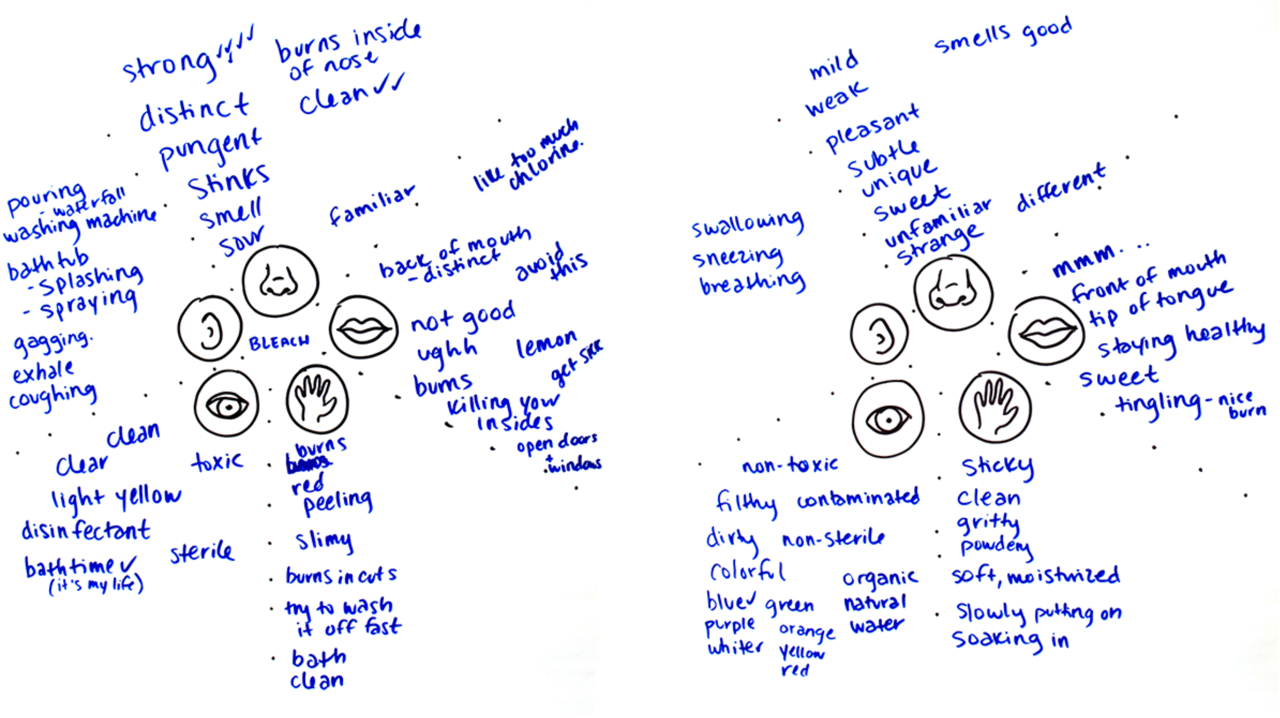
As the advisors had misconceptions about MRSA, the team developed a summary of basic information about MRSA to help educate MEDiC study participants. The summary is included in the tracking booklets and includes information about what MRSA is, who is most at risk of MRSA infections, and how MRSA is spread. In addition, the kit's tracking booklets included answers to common questions the advisors asked about MRSA. In particular, they answer many of the questions about what activities a person with MRSA should avoid during an outbreak, as this was a major concern and knowledge gap for the advisor group.

Bleach Has Positive and Negative Connotations

What We Heard

The Five Senses map for bleach (Figure 2) was not entirely negative. Bleach was perceived as clean and sterile, a tool for disinfecting and ensuring a safe environment. Many of the advisors had already utilized bleach bath protocols for their children, so bleach was also associated with familiarity and bath time. This is bolstered by the opposite Five Senses map (Figure 2), which includes words such as “dirty” and “contaminated.” Despite the positive associations, bleach was also associated with many negative senses. It was seen as something that would cause negative effects such as burning the skin, nose, and eyes and causing gagging. One advisor associated the sense of taste of bleach with “killing your insides” because it was toxic to consume. On the opposite Five Senses map, the advisors used words such as “pleasant,” “sweet,” “subtle,” and “tingling” to describe the opposite of bleach.

Figure 2. Five Senses map of bleach and the opposite of bleach.



What We Did

Team members DL and CM, who have expertise in visual communication design, created a final Five Senses map for the MEDiC study itself that incorporates the positive aspects of the Five Senses map of bleach and the opposite Five Senses map of bleach (Figure 2). This final map (Figure 3) guided branding and design efforts. The idea of utilizing bleach baths, particularly for children, understandably produces anxiety for some parents. The Five Senses map of bleach shows that bleach is very stimulating; therefore, the goal for visual communication was to provide a more mild, low-impact backdrop for the study by being visually well-organized with low visual stimulation.

A final logo was created (Figure 4). The logo represents both a water droplet and the shape of MRSA under a microscope and uses cool colors as recommended by the Five Senses map. In addition, an Indiana University logo was included in all of the materials to lend credibility to the content.

The kit uses photographs to show MEDiC study participants what the materials are and how they are used. To keep the designed materials light, clean, soft, etc, the page layout is simple with an abundance of white space. The chosen paper stock is bright white, and the text is gray rather than a visually harsher black. The covers of the tracking booklets are a plain pattern so that MEDiC study participants can carry them without signaling to strangers that they are in an MRSA study.

Figure 3. Five Senses map of the Methicillin-Resistant Staphylococcus aureus Eradication and Decolonization in Children study.

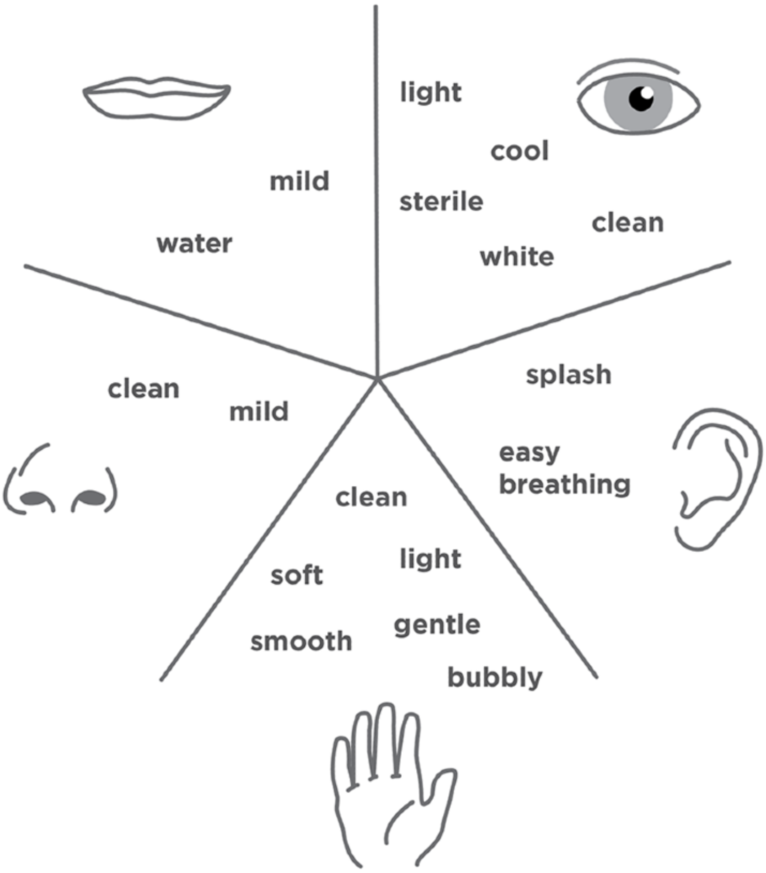


Figure 4. The Methicillin-Resistant Staphylococcus aureus Eradication and Decolonization in Children study logo.

The Final Kit Components

The following is a summarized list of each of the final components of the MEDiC kit, which can be seen in Figure 5. The MEDiC kit is available for download on the Research Jam website [30]. The MEDiC trial randomizes participants into 2 arms: a decolonization arm and a hygiene-only arm. Some of the kit components were given to one arm or the other or to both. This is indicated in parentheses in the descriptions that follow.

Figure 5. The Methicillin-Resistant Staphylococcus aureus Eradication and Decolonization in Children study kit components.

1. *Decolonization instruction booklet (decolonization*; Multimedia Appendix 4): This booklet contains detailed step-by-step photographic instructions for preparing and completing bleach baths and mupirocin swabs and tracking them with the decolonization tracking booklet. The instruction booklet also includes a link to the bleach bath instructional video on the Web.
2. *Decolonization instructional video (decolonization*; Multimedia Appendix 5): This is a short video showing the same step-by-step information included in the decolonization instruction booklet for both the bleach baths and the mupirocin swabs. The demonstration was performed by a pediatrician from Riley Hospital for Children (AC) to ensure that the MEDiC study participants know the information is reliable.
3. *Decolonization tracking booklet (decolonization*; Multimedia Appendix 7): A booklet to help families who are randomized into the decolonization group track hygiene and decolonization tasks as well as MRSA-related outcomes. The book begins with information about MRSA to help patients and families understand better how MRSA happens and to answer key questions advisors in our group asked. This tool aids patients and their families in keeping track of the MRSA hygiene and decolonization tasks they complete, making it easier for MEDiC study participants and more accurate for researchers.
4. *Packing slip (decolonization*; Multimedia Appendix 6): A photographic list of decolonization supplies sent to families randomized to the decolonization group. The supplies include a 3-gallon bucket, a wax crayon, a small measuring cup, a large measuring cup, *Splash-less* bleach, cotton swabs, mupirocin, a hygiene shower hanger, a decolonization tracking booklet, and a decolonization instruction booklet.
5. *Hygiene shower hanger (both*; Multimedia Appendix 9): A shower hanger with reminders for good MRSA hygiene (to be printed and laminated before use). This helps patients and families remember key things they should be doing for the hygiene portion of the protocol and is located in a place where they will see it every day: the shower.
6. *Hygiene tracking booklet (hygiene only*; Multimedia Appendix 8): A booklet to help families who are randomized into the hygiene-only group track MRSA hygiene tasks and MRSA-related outcomes. The book begins with information about MRSA to help patients and families understand better how MRSA happens and to answer key questions advisors in our group asked. This tool aids patients and their families in keeping track of the MRSA hygiene activities they complete, making it easier

for MEDiC study participants and more accurate for researchers.

7. *Decolonization supplies (decolonization*; Multimedia Appendix 4 gives more information on how the supplies are used): A bucket, a large measuring cup, a small measuring cup, a wax crayon, *Splash-less* bleach, cotton swabs, and mupirocin ointment.

Follow-up

In total, 5 parent advisors participated in a small follow-up survey in which they were asked to provide feedback on the MEDiC kit. Overall, the advisors had positive reactions to the kit, finding it more helpful than other materials they had been given in the past for MRSA. In particular, they found the instruction booklet to be very helpful. They also provided some suggestions for further improvement, such as including tips for helping to ease discomfort from MRSA or instruction on how to properly clean the bathtub after bleach baths. This feedback suggests that the kit is promising as a tool for helping families with MRSA decolonization even in its current state, but that additional refinements are likely to be found with use and further assessment.

Discussion

Contribution to the Literature

In preparation for the MEDiC study, our randomized controlled trial, we engaged 16 advisors (adolescent patients along with parents) with previous experiences with MRSA infections requiring abscess I&D. We engaged them primarily around the concept of hygiene instructions as well as the procedures for MRSA decolonization with the ultimate goal of preventing recurrent infections. These key stakeholders provided us with the perspective of patients with lived experiences and the feedback necessary to develop an MRSA decolonization toolkit for use in the MEDiC study.

Our advisors identified a number of barriers/pain points regarding the 12 MRSA hygiene and decolonization tasks. From these, we were able to extract 5 major themes (1) existing step-by-step decolonization instructions are not user friendly and do not answer advisors' common questions; (2) families may not have the proper tools for implementing decolonization and hygiene procedures; (3) some decolonization and hygiene procedures may be out of the control of families; (4) decolonization and hygiene procedures are burdensome; (5) advisors had incorrect assumptions about MRSA despite having experience with it; and (6) bleach has positive and negative connotations. In addition, the advisors were able to brainstorm potential solutions for these identified pain points. Building upon the work of the advisors during the workshop, we designed the MRSA decolonization kit. The kit includes (1) an instruction booklet with step-by-step photographic instructions, (2) a bleach bath and mupirocin swab instructional video, (3) a decolonization and hygiene tracking booklet with MRSA education, (4) a packing slip with a list of all study materials, (5) an MRSA hygiene shower hanger as an easily accessible reminder of the hygiene steps, (6) a hygiene tracking booklet with MRSA education, and (7) decolonization supplies (including a bucket, measuring cups, and bleach) required for the decolonization protocol.

This patient-engaged approach is fairly new to MRSA. We acknowledge that the individual hygiene steps and decolonization procedures themselves are not novel and have been well documented [11,15,16,31,32]. However, this *kit*, which incorporates the feedback of patient partners, is—as far as we can tell—the first documented attempt to create a detailed, user-friendly approach to MRSA home treatment based in part on patient engagement. Furthermore, our study is the first engagement effort to include primarily a pediatric population and their parents regarding the topic of MRSA decolonization. In fact, to our knowledge, this represents one of only a handful of published studies regarding patient engagement on the topic of community-acquired MRSA. For instance, a translational research collaborative in New York City recruited and trained barbers and hair stylists from 9 barbershops as part of community engagement and education efforts to create awareness of the dangers of MRSA infections [33]. That, however, was engagement in the implementation phase rather than our approach, which brought in patient partners in the study design phase. Patients as design partners bring a unique perspective to traditional research projects, which is among the reasons this approach is championed by funding organizations such as the Agency for Healthcare Research and Quality (AHRQ) and the Patient-Centered Outcomes Research Institute [34].

The Role of Human-Centered Design

HCD is a helpful tool for patient engagement and is especially relevant in instances when product design is performed. A study intervention tool (such as the MEDiC kit) is a product that can be used by individuals within a study. HCD has a rich tradition of working to ensure that products are designed with good *fit* for users—patients and parents in this case [25]. Through the participatory methods in the workshop, the advisors were empowered to begin the design process by thinking through the implementation barriers to be solved by the design and potential solutions to these. Facilitators CM and DL acted as design process experts, guiding the advisors through activities that helped them act as co-designers who set the direction and parameters for the design itself.

HCD analysis is qualitative and, as such, employs many different methods across different projects. For this study, authors CM and DL employed Kolko's analysis strategy based on the DIKW framework. The DIKW framework itself was developed in the field of computer systems. Criticisms of this framework include, first, that it defines the words used for each stage vaguely or in contrast to preexisting definitions. Second, the visual representations of the framework (particularly as a pyramid) imply that each stage can be reached by filtering the content of the previous stage and that this misrepresents the more complicated relationships between data, information, knowledge, and wisdom. Kolko's work addresses these criticisms in that he defines the concepts in useful terms (particularly in the context of design), describes the gaps between stages, and proposes methods that employ human reasoning and action while processing content in each stage to jump the next gap

[29]. As Kolko explains, this process is not as linear as it might be described or perceived. The movement between stages in DIKW is iterative and based not on procedure but on human judgment. Ultimately, there is an interaction between the content of each DIKW stage, the context of the problem to be solved, and the experience each team member brings to their processing of the content. The end stages of DIKW in particular require making *best guesses* based on what was known previously, what has been previously experienced, and new information gained from the data. This is what the HCD discipline often refers to as *intuition*, but is essentially the connectionist view of cognition, which describes what the human brain does to create new concepts where no clear model exists [35]. Kolko describes this in terms of abductive reasoning, in which one produces plausible conclusions based on related knowledge and experiences and some inference [29]. There is an important role for intuition in design, but this *intuition* or connectionist cognition must be grounded in the data. Kolko's use of DIKW is helpful for providing a structure for analysis and synthesis, and ensuring intuition is led by the data and applied at appropriate stages in the process.

The Role of Visual Communication

Visual communication strategies, such as those from sensory marketing, must be undertaken with care. The advisors did comment on this during the discussion of changing bleach to be more appealing. In particular, they discussed that they would not want bleach to be changed in a way that would make it look like something a child would want to drink. They wanted the visual communication aspects signaling bleach's toxicity to remain. The study team did not intend to repackage bleach but did want to avoid unintended amplification of concerns MEDiC study participants might have about bleach. As discussed above, bleach is a substance with many negative associations that are quite reasonable. Given this, the team wanted the materials and language for the study to avoid adding to the understandable anxiety some potential participants might feel when considering a protocol involving bleach baths, particularly for their children. The goal was not to persuade participants but to ensure that the materials did not act as an additional layer of stress or confusion. Visual communication and messaging can be very powerful tools that can cause harm, such as the tobacco industry targeting underage youth to increase tobacco use [36]. On the contrary, visual communication and messaging have been used to improve health behaviors, as in the case of the truth campaign's successful antismoking advertisements targeting young people [37]. In the context of this particular research study, though the team was designing materials for a protocol believed to be beneficial, the team was careful to focus on clarity, ease of use, and low stimulation rather than persuasion. The team wanted MEDiC study participants to have the opportunity to participate while fully informed and with as smooth a process as we could provide. The team is hopeful for future opportunities to test and refine the kit within the context of a large study as originally planned.

Limitations

One limitation is the small sample size of the advisors who participated in the workshop. However, all of the patients and parents had experience with skin infections such as MRSA, and a few had previously or were currently utilizing bleach bath protocols, thus making them highly knowledgeable stakeholders in the problem we were trying to solve. Additionally, the team recruited parents with children ranging from 15 months to 18 years of age and adolescents ranging from 10 to 18 years of age attended the workshop. This allowed for a range of perspectives based on patient age.

Another limitation is that we were unfortunately unable to schedule a second workshop that would have worked for the advisors between completion of the kit and the study start date. Thus, only 1 session was held, leaving open the possibility that subsequent sessions might have identified other barriers or additional refinements to the design of the kit. HCD often preaches a *fail early and often* approach, valuing iterative cycles; what Hassi and Laakso call *thinking by doing* [38]. In this spirit, rather than a longer research process to create the *perfect* kit the first time (and risking being off the mark), the team planned to iteratively engage MEDiC trial participants after they began using the prototype kit to explore additional barriers that may have emerged and to identify additional kit refinements that might be made. Unfortunately, due to recruitment challenges in the MEDiC trial, there were too few active participants to engage meaningfully in this effort. We still feel that the kit would benefit from iterative refinement in future work.

Conclusions

In this first documented attempt to incorporate pediatric patients and their families as key stakeholders regarding MRSA SSTIs, we engaged advisors in an HCD process to cocreate a toolkit to help participants complete MRSA decolonization and hygiene protocols as part of a comparative effectiveness trial comparing hygiene-only education versus decolonization protocols on infection recurrence. From the perspective of patients with lived experiences, these advisors provided the study team with a better understanding of the potential barriers to completion of study protocols the participants would likely face as well as guidance on the design of the kit. This stakeholder engagement was essential and directly led to the development of an MRSA decolonization toolkit (MEDiC kit), which was implemented in the MEDiC comparative effectiveness trial.

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Conflicts of Interest

PM has received research funding from Trevena Inc, the Emergency Medicine Foundation, and the Society of Academic Emergency Medicine Foundation.

Multimedia Appendix 1
Task analysis grid 2.
[PNG File , 66 KB - jopm_v12i2e14974_app1.png]

Multimedia Appendix 2
Task analysis grid 3.
[PNG File , 92 KB - jopm_v12i2e14974_app2.png]

Multimedia Appendix 3
Task analysis grid 4.
[PNG File , 58 KB - jopm_v12i2e14974_app3.png]

Multimedia Appendix 4
Decolonization instruction booklet.
[PDF File (Adobe PDF File), 4918 KB - jopm_v12i2e14974_app4.pdf]

Multimedia Appendix 5
Decolonization instructional video.
[MP4 File (MP4 Video), 18879 KB - jopm_v12i2e14974_app5.mp4]

Multimedia Appendix 6
Packing slip.
[PDF File (Adobe PDF File), 1509 KB - jopm_v12i2e14974_app6.pdf]

Multimedia Appendix 7
Decolonization tracking booklet.
[PDF File (Adobe PDF File), 157 KB - jopm_v12i2e14974_app7.pdf]

Multimedia Appendix 8
Hygiene tracking booklet.
[PDF File (Adobe PDF File), 141 KB - jopm_v12i2e14974_app8.pdf]

Multimedia Appendix 9
Hygiene shower hanger.
[PDF File (Adobe PDF File), 92 KB - jopm_v12i2e14974_app9.pdf]

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Abbreviations

AHRQ: Agency for Healthcare Research and Quality
DIKW: data, information, knowledge, wisdom
HCD: human-centered design
I&D: incision and drainage
MEDiC: methicillin-resistant Staphylococcus aureus Eradication and Decolonization in Children
MRSA: methicillin-resistant Staphylococcus aureus
RJ: Research Jam
SSTI: skin and soft tissue infection

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Methicillin-Resistant Staphylococcus aureus Eradication and Decolonization in Children Study (Part 2): Patient- and Parent-Centered Outcomes of Decolonization

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Abstract

Background: Skin and soft tissue infections (SSTIs) due to community-acquired methicillin-resistant *Staphylococcus aureus* (MRSA) can lead to a number of significant known medical outcomes including hospitalization, surgical procedures such as incision and drainage (I&D), and the need for decolonization procedures to remove the bacteria from the skin and nose and prevent recurrent infection. Little research has been done to understand patient and caregiver-centered outcomes associated with the successful treatment of MRSA infection.

Objective: This study aimed to uncover MRSA decolonization outcomes that are important to patients and their parents in order to create a set of prototype measures for use in the MRSA Eradication and Decolonization in Children (MEDiC) study.

Methods: A 4-hour, human-centered design (HCD) workshop was held with 5 adolescents (aged 10-18 years) who had experienced an I&D procedure and 11 parents of children who had experienced an I&D procedure. The workshop explored the patient and family experience with skin infection to uncover patient-centered outcomes of MRSA treatment. The research team analyzed the audio and artifacts created during the workshop and coded for thematic similarity. The final themes represent patient-centered outcome domains to be measured in the MEDiC comparative effectiveness trial.

Results: The workshop identified 9 outcomes of importance to patients and their parents: fewer MRSA outbreaks, improved emotional health, improved self-perception, decreased social stigma, increased amount of free time, increased control over free time, fewer days of school or work missed, decreased physical pain and discomfort, and decreased financial burden.

Conclusions: This study represents an innovative HCD approach to engaging patients and families with lived experience with MRSA SSTIs in the study design and trial development to determine meaningful patient-centered outcomes. We were able to identify 9 major recurrent themes. These themes were used to develop the primary and secondary outcome measures for MEDiC, a prospectively enrolling comparative effectiveness trial.

Trial Registration: ClinicalTrials.gov NCT02127658; https://clinicaltrials.gov/ct2/show/NCT02127658

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KEYWORDS

patient-centered outcomes; Staphylococcus aureus; abscess; decolonization; human-centered design

Introduction

Background

Community-acquired skin and soft tissue infections (SSTIs) such as cellulitis, boils, myositis, and abscesses caused by antibiotic-resistant bacteria known as methicillin-resistant *Staphylococcus aureus* (MRSA) have risen dramatically over the past 20 years, and a significant proportion of these affect otherwise healthy children [1-8]. The estimated incidence of hospitalizations due to MRSA SSTIs is more than 45 per 100,000 children, with many children requiring surgical procedures such as incision and drainage (I&D), to drain pus caused by the infection [6,9,10]. Even with appropriate treatment, the rate of recurrent infection can be as high as 72% [11-16]. Thus, strategies are needed to reduce the rate of recurrent infection and comorbid suffering, cost, and health care utilization. One strategy, called *decolonization*, focuses on eradicating the presence of bacteria on the skin and in the nose of people at risk of infection. Examples of accepted decolonization protocols include the use of topical mupirocin (antibiotic) ointment in the nose to eliminate nasal carriage and chlorhexidine or bleach baths to eliminate skin carriage [17-19].

Objectives

The MRSA Eradication and Decolonization in Children (MEDiC) comparative effectiveness trial [20] aimed to assess the effectiveness of 2 interventions: (1) abscess surgery and hygiene education compared with (2) abscess surgery and hygiene education followed by decolonization. Along with clinical measures, the study team aimed to understand the effects of these interventions on patient-centered outcomes. However, during the planning phase of our study, literature on patient-centered outcome measures for MRSA infection treatment was very limited. To uncover patient-centered outcomes that might be measured, we engaged patients with lived experiences with MRSA SSTIs and their caregivers in a human-centered design (HCD) workshop. This paper will discuss the workshop activities and results and how we incorporated these patient-centered outcomes into our overall study. The authors do not see this work as a final product, but rather a first step in inspiring the creation of a comprehensive set of MRSA patient-centered outcomes that can be measured alongside clinical outcomes in future work. A companion paper discusses a separate objective of this project, which was to engage patients and their families in the design of an MRSA decolonization toolkit to support families in the MEDiC study in adhering to the decolonization process [21].

Methods

Overview

The Indiana University School of Medicine Institutional Review Board approved this study. Participants were invited to participate in the workshop if they were a parent of a patient (3 months-18 years) who had undergone an I&D procedure at Riley Hospital for Children or were a patient (9-18 years) who had undergone an I&D procedure. All study participants (advisors) provided written informed consent, and children aged 9 to 13 years provided assent. All participants received US $20 per hour for their time and participation. The team hosted a 4-hour workshop that was audio-recorded.

Human-Centered Design

HCD utilizes participatory methods of engagement to empower people to share their experiences, express their thoughts, and generate new ideas through what they say, do, and make [22]. *Say* methods include guided discussion, interviews, or questionnaires and elicit explicit information. *Do* methods include observation and elicit information that can be directly viewed. *Make* methods include collage, drawing, and 3D modeling and—through the maker's explanation of their creation—elicit tacit information (or information that is known but difficult to access and express). When used in combination, these participatory methods help to ensure valuable involvement and can uncover the unmet needs of stakeholders [22]. Research Jam (RJ), the patient engagement core of the Indiana Clinical and Translational Sciences Institute, applies an HCD approach in the context of health research, collaborating with principal investigators such as the MEDiC study's PM. HCD was chosen because its participatory methods— *make* methods in particular—are useful in helping participants create symbolic expressions of their tacit knowledge about the topic and provide a scaffolding from which participants can speak about their experiences. Patient and parent-centered outcomes, particularly those that are not immediately observable, but are *below the surface* are by their nature tacit. The study team was particularly careful to include activities that made space for the adolescents, who might feel uncomfortable speaking up in the more discussion-based portions of a workshop filled with adults. Activities that were first completed independently and then shared with the rest of the group allowed all participants to respond and share equally. This is important because research shows that parent reporting of adolescent outcomes may differ from self-reported adolescent outcomes, meaning that parent reporting ought not to be substituted for self-reporting [23]. In addition, we wanted to understand the personal experiences of the parent advisors, as we assumed MRSA affects them differently than it does their children. In this spirit, we aimed to gather self-reported experiences from adolescent advisors as well as the unique self-reported experiences of parent advisors themselves (as opposed to their interpretation of their children's experiences). Participatory HCD methods were used to help both patient and parent advisors express unearthed outcomes of MRSA decolonization in an effort to, ultimately, create a patient-centered outcomes measurement tool.

The Workshop

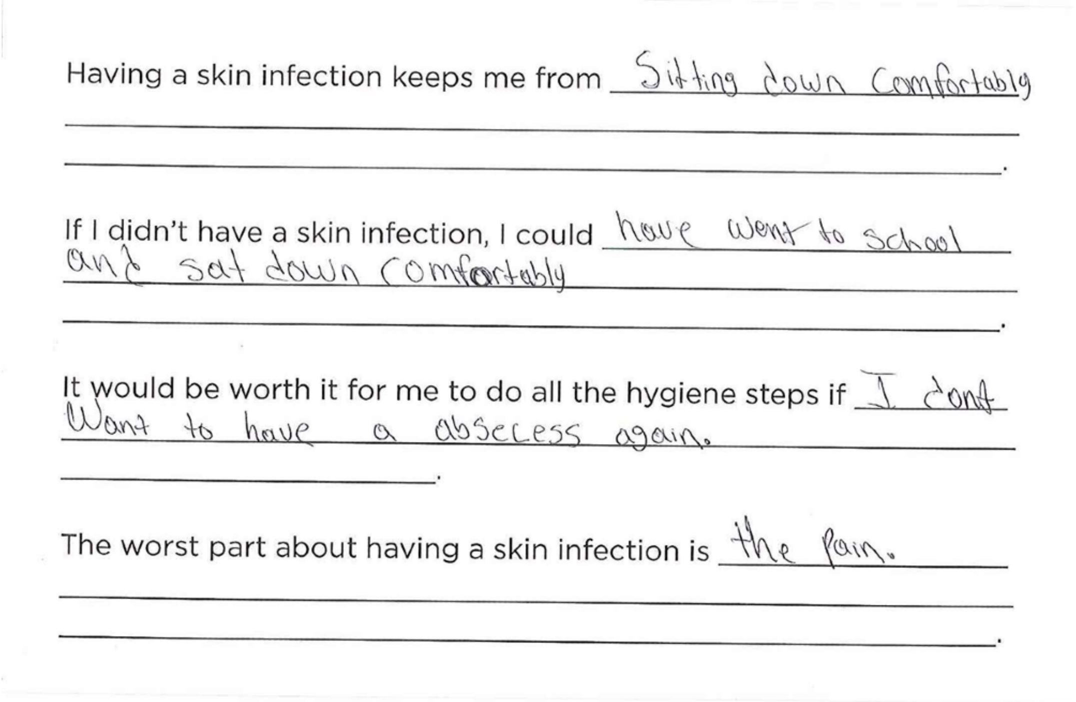
The research team held a 4-hour workshop (with a 30-min break for lunch) to explore a series of topics. One of these was the patient- and parent-centered outcomes of MRSA treatment. A total of 5 patient advisors who had undergone an I&D procedure attended the workshop. There were 3 females aged 10, 14, and 18 years and 2 males aged 14 and 18 years. In total, 11 parent advisors attended the workshop. This included 4 female parents of the adolescents (1 parent brought 2 children) and 7 additional parents (6 females and 1 male) of younger children (aged 15 months to 5 years) who had also experienced I&D. The patient advisors remained with their parents throughout the workshop.

The 2 activities, *Fill-in-the-blank* and *Collage*, were utilized to gain an understanding of the outcomes of importance to patients and parents.

Fill-in-the-Blank

Fill-in-the-blank is a *say* method that utilizes writing. Advisors were given a worksheet (Figure 1) with 4 fill-in-the-blank statements. The worksheets included the following statements:

Figure 1. Patient advisor fill-in-the-blank discussing discomfort from methicillin-resistant Staphylococcus aureus.



Having a skin infection keeps me from Sitting down Comfortably

If I didn't have a skin infection, I could have went to school and sat down comfortably

It would be worth it for me to do all the hygiene steps if I dont Want to have a absecess again.

The worst part about having a skin infection is the pain.

1. Having a skin infection keeps me from ______________.
2. If I didn't have a skin infection, I could ______________.
3. It would be worth it for me to do all the hygiene steps if ______________.
4. The worst part about having a skin infection is ______________.

Parent worksheets included the following statements:

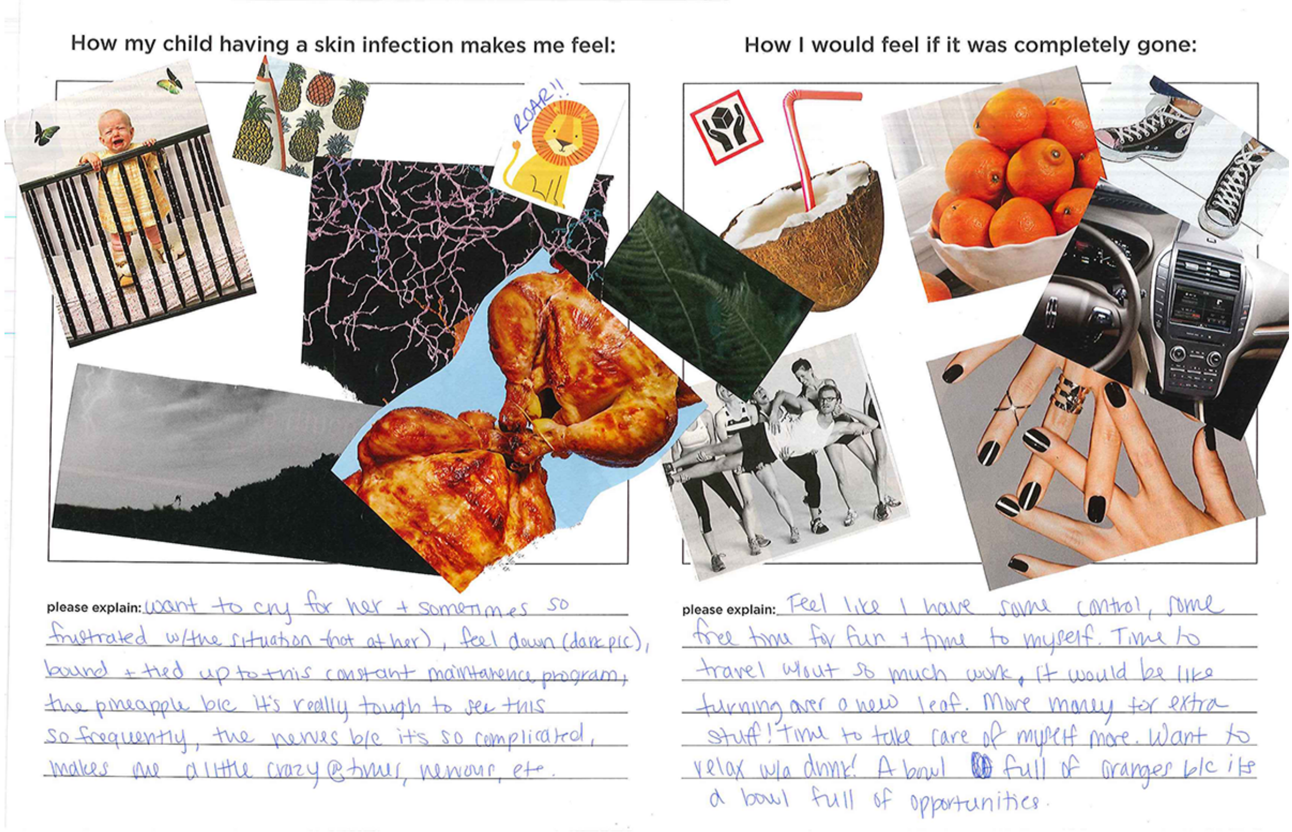
1. Having a skin infection keeps my child from ____________.
2. My child having a skin infection keeps me from ______________.
3. If my child didn't have a skin infection, we could ______________.
4. It would be worth it for me to do all the hygiene steps if ____________.

Facilitators asked patient and parent advisors to share their answers and prompted follow-up questions to encourage discussion within the group.

Collage

Collage is a *make* method in which the participant is asked to express their thoughts and feelings using images rather than strictly words. This approach allows participants to express themselves where words may fail, it aids in common understanding through the utilization of symbols and metaphors and reveals relationships that are harder to uncover through verbal or written presentations [24]. Participants are first given a large and varied selection of images not directly related to the topic of discussion and asked to create a collage that responds to a given prompt. They are then asked to write a brief explanation of the images they chose and how they relate to the topic of interest. Finally, they are asked to show their collage to the group and explain aloud why they chose the images they chose. The group is asked to respond to each participant by talking about parts of each collage they can relate to. During the workshop, a large selection of images, including both abstract and representative images, was placed in a pile in the center of the table. Advisors were given a large worksheet including 2 different prompts with empty space below. Each prompt asked the advisor to create a collage by gluing images of their choosing in the spaces provided. Below each collage space, blank lines were provided for the advisor to explain their collages (Figure 2). Patient advisors were given the prompts: *How having a skin infection makes me feel* and *How I would feel if it was completely gone*. Parent advisors were given the prompts: *How my child having a skin infection makes me feel* and *How I would feel if it was completely gone*. Facilitators asked advisors to share their worksheets with the group and prompted follow-up questions to encourage discussion within the group.

Figure 2. Parent advisor collage.



How my child having a skin infection makes me feel:

please explain: want to cry for her + sometimes so frustrated w/the situation (not at her), feel down (dark pic), bound + tied up to this constant maintanence program, the pineapple b/c it's really tough to see this so frequently, the nerves b/c it's so complicated, makes me a little crazy @ times, nervour, etc.

How I would feel if it was completely gone:

please explain: Feel like I have some control, some free time for fun + time to myself. Time to travel w/out so much work, it would be like turning over a new leaf. More money for extra stuff! Time to take care of myself more. Want to relax w/a drink! A bowl full of oranges b/c its a bowl full of opportunities.

Analysis

The analysis process for this research was inductive in nature and, specifically, based on Kolko's methods for using the data, information, knowledge, wisdom framework, which describes the steps by which data (discrete symbols) are processed in stages to, ultimately, reach wisdom (development of increased value) [25]. As the ultimate goal of this research was to describe patient-centered outcome domains, the level of processing of our data stops at the knowledge stage. Responses from the fill-in-the-blank and collage worksheets were transcribed, and discrete ideas were separated into individual snippets of information. Additionally, discrete ideas from the audio-recorded discussion that added additional details were transcribed and added as individual snippets. In total, 2 members of the research team (CM and DL) who attended the workshop utilized affinity clustering to collaboratively organize the snippets. This method is an iterative process for grouping data by relationship (typically similarity) to move from data to information [25]. Once revisions to the clusters were finished, each cluster was then reviewed for content, discussed, and given a name to represent the theme of the snippets it contained. From here, a descriptive phrase was created for each theme to communicate its meaning to others.

Results

Workshop Participation

In total, 16 advisors attended the workshop, and 5 of these were adolescents (3 females aged 10, 14, and 18 years; 2 males aged 14 and 17 years). In total, 11 parents attended (10 females and 1 male; 4 parents of the adolescents and 7 parents of children aged 15 months to 5 years). Adolescents and their parents were kept together during the workshop.

Key Themes

The following are the key themes related to outcomes of importance to patients with MRSA and their parents. Some of the key themes were mentioned by patient advisors, some by parent advisors, and many by both. This is indicated in brackets next to the theme in the descriptions below. This does not indicate importance. For example, although parent advisors did not report pain or discomfort as often as patient advisors, this does not indicate that their child's discomfort is unimportant. Parent advisors were often explicitly asked to report about their own perspectives, emotions, and needs. The fill-in-the-blank worksheet, for example, asked about how their child's skin infection limited their child as well as how it limited them. The collage worksheet asked about their own feelings rather than their child's feelings. Table 1 shows an example of the process by which the team arrived at these themes.

Table 1. Process to move from quotes to cluster to theme.

Exemplary quotes	Cluster	Theme
• "Having a skin infection keeps me from showing off too much of my skin." • "If my skin infection was gone, I'd feel beautiful…" • "Having a skin infection makes me feel old."	• MRSA[a] keeps me from feeling attractive and healthy	• Improved self-perception
• "Having a skin infection keeps my child from being their own self." • "If I didn't have a skin infection, I could wear what I want."	• MRSA keeps the patient from being his or her self	• Improved self-perception
• "Older kids may not want to join sports things because of the worry. It would give them the freedom to say, 'Hey, I want to go out for this sports team and I'm not going to be scared that I'll have an outbreak and I'm not going to be able to play or people will think I'm weird because of this infection.'" • "Having a skin infection keeps my child from feeling like the rest [of the girls at ballet] – having to wear [long sleeved leotards to cover her infection when the rest of the girls wear short sleeves]."	• MRSA makes my child feel out of place	• Improved self-perception

[a] MRSA: methicillin-resistant *Staphylococcus aureus.*

Fewer Methicillin-Resistant Staphylococcus aureus Outbreaks (Patient and Parent Advisors)

Patient and parent advisors ultimately wanted to experience fewer skin infection outbreaks. This was a very common answer for the question: *It would be worth it for me to do all the hygiene steps if* _________. Some advisors reported that they would do all of the hygiene steps for any degree of improvement, whereas others thought it would be worth it if they had no more outbreaks at all (Figure 1).

Decreased Physical Pain and Discomfort (Patient Advisors)

The patient advisors wrote and talked about the discomfort they felt from their skin infections. One advisor wrote: "Having a skin infection keeps me from sitting down comfortably" (Figure 1). Other patient advisors mentioned episodes of intense itching. Parent advisors whose children were too young to put their discomfort into words were very interested in hearing from the patient advisors about what their skin infections felt like as this gave them some idea of how much discomfort their own children might be experiencing.

Improved Emotional Health (Patient and Parent Advisors)

Outbreaks from skin infections cause negative emotions such as stress, anger, and sadness. One patient advisor wrote about his collage: "[Having a skin infection] makes me feel like I wanna blow up."

One parent wrote about her collage: "[I chose the image of nerves] because it's so complicated, makes me a little crazy at times, nervous, etc." Getting rid of the infection would result in "Simple peace of mind…because if we see a spot, we're just stressed." Parent advisors reported that they were constantly alert for new spots that could potentially lead to another long course of treatment: "One little pimple is not one little pimple. It could turn into a full-blown softball and then we're down at the hospital." In many of the collages, advisors wrote that if their skin infections were completely gone, they would feel calm, relaxed, or happy. As one parent wrote in the fill-in-the-blank activity: "[If my child didn't have a skin infection, we] wouldn't be scared of all the things we can't control." (Figure 3).

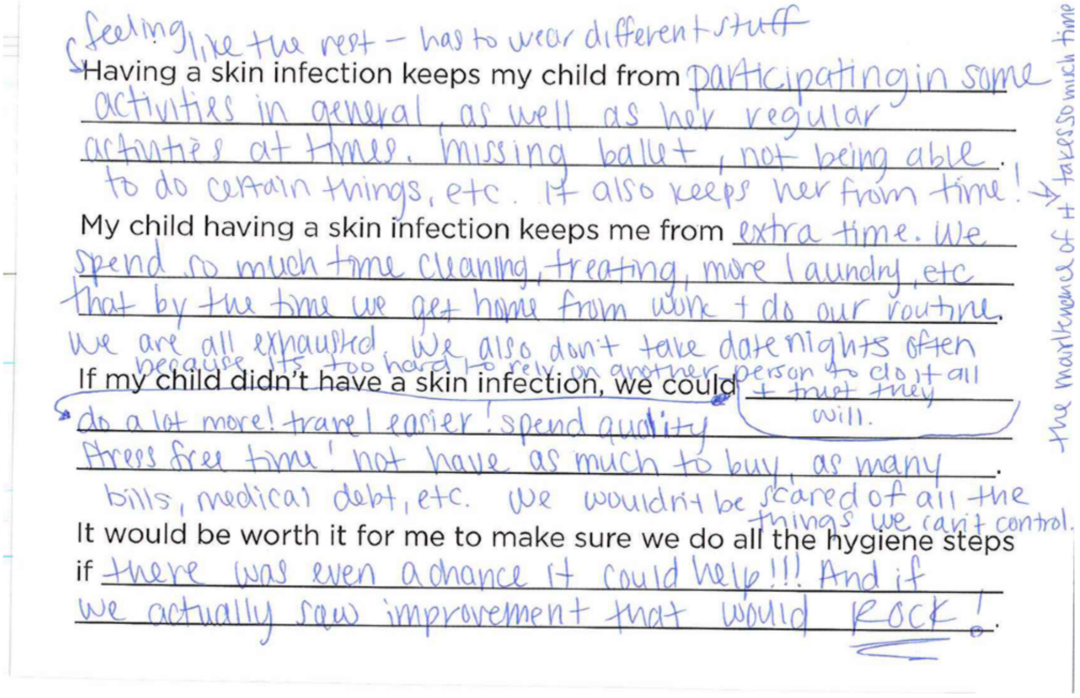


Figure 3. Parent advisor fill-in-the-blank.

feeling like the rest – has to wear different stuff

Having a skin infection keeps my child from participating in some activities in general, as well as her regular activities at times. missing ballet, not being able to do certain things, etc. It also keeps her from time! → the maintenance of it takes so much time

My child having a skin infection keeps me from extra time. We spend so much time cleaning, treating, more laundry, etc that by the time we get home from work + do our routine. We are all exhausted. We also don't take date nights often because its too hard to rely on another person to do it all + trust they will.

If my child didn't have a skin infection, we could do a lot more! travel easier! spend quality stress free time! not have as much to buy, as many bills, medical debt, etc. We wouldn't be scared of all the things we can't control.

It would be worth it for me to make sure we do all the hygiene steps if there was even a chance it could help!!! And if we actually saw improvement that would ROCK!

Improved Self-Perception (Patient and Parent Advisors)

Patient advisors mentioned that having a skin infection made them feel "dirty" or "not clean." One patient advisor wrote in her fill-in-the-blank activity about how her skin infection affected her ability to express herself through her appearance:

> *[Having a skin infection] keeps me from showing off too much of my skin. [If I didn't have a skin infection, I could] wear what I want and do what I want.*

In her collage activity, she wrote about her skin infection's effect on how she sees herself:

> *When having a skin infection, I feel…old. When the skin infection's completely gone, I feel beautiful.*

One mother talked about how her daughter stood out at ballet because she wore a long sleeve leotard to cover her skin infection whereas the other girls all wore short sleeves.

Decreased Social Stigma (Patient and Parent Advisors)

Patient and parent advisors described stigma associated with skin infections such as MRSA. As 1 parent advisor explained:

> *Older kids may not want to join sports things because of the worry. It would give them the freedom to say, "Hey, I want to go out for this sports team and I'm not going to be scared that I'll have an outbreak and…people will think I'm weird because of this infection."*

Stigma did not only affect the patients but also their parents. One parent advisor shared a story about discovering, just days after hosting children for her daughter's birthday party, that her daughter had MRSA. When she contacted the parents of the other children who attended to let them know, a few were angry and asked her why she had planned a party when her daughter had MRSA. She felt that these other parents were accusing her of being negligent, but she insisted that she would never have exposed anyone's children to MRSA had she realized that her daughter was infected.

Increased Amount of Free Time (Parent Advisors)

Managing a skin infection such as MRSA takes up a lot of time. Parent advisors especially commented on the lack of free time they experienced due to all of the cleaning, sanitizing, laundry, visits to the doctor, trips to the emergency department, etc. One parent advisor wrote about her collage:

> *[If the infection were gone, I would feel] like I have some free time for fun and time to myself…[I chose the image of] a bowl full of oranges because [my child not having a skin infection would be] a bowl full of opportunities.*

Increased Control Over Free Time (Patient and Parent Advisors)

Patient and parent advisors reported that MRSA keeps them from participating in activities they enjoy. As 1 parent advisor explained in the fill-in-the-blank activity: "[Having a skin infection keeps my child from] participating in her regular activities at times [like] missing ballet." In particular, parent advisors frequently mentioned wanting to be able to take their children to public pools but feeling that they could not because of the risk of their child infecting others. There was a lively discussion about whether or not chlorine in pools would keep the infection from spreading to others:

> *I've got a pool and we're going to put it up, but it'll have chlorine in it…I mean chlorine is bleach right? Does that help? Because that would determine whether or not we let other kids come over to swim in our pool.*

Discomfort during an outbreak also kept some of the children from going to school or playing with friends and siblings. In addition, 1 married couple from the parent advisor group reported that the difficulty of explaining MRSA protocols to babysitters had kept them from going on their usual date nights as a couple: "We don't go on date nights often because it's too hard to rely on another person to do [the whole routine]." Traveling, in general, was something many families avoided while they were implementing decolonization protocols. To complete bleach baths during a vacation, for example, families would need to ensure there would be a bathtub available where they were staying and would need to bring bleach and measuring equipment (or purchase these upon arrival). The complications of travel during decolonization led many families to avoid it entirely.

Fewer Days of School or Work Missed (Patient and Parent Advisors)

Advisors reported that they sometimes had to miss school and work due to skin infection outbreaks. The patient advisors discussed having to miss school because their skin infection outbreaks made them too uncomfortable to concentrate. Parent advisors discussed missing work and their child missing school to receive medical care for outbreaks.

Decreased Financial Burden (Parent Advisors)

Parent advisors reported that caring for MRSA infections takes a lot of financial resources. There are medical bills from visits to the doctor, I&D procedures, emergency department visits, and prescriptions. In addition, there are added costs for purchasing bleach and other supplies and increased bills for water and electricity from extra baths and laundry. These additional costs are compounded by lost wages from parents missing work to take their children to receive care during work hours. These costs add up quickly and create additional burdens on families. One parent advisor wrote in her fill-in-the-blank activity: "If my child didn't have a skin infection, we could not have as much to buy, as many bills, medical debt, etc." (Figure 3).

Discussion

Development of Patient-Centered Outcome Measures

Outcomes research is increasingly incorporating the patient's perspective in the study design and development of outcome measures [26-29]. This is thought to improve the credibility of research results and can be considered an ethical imperative [30]. Although the effectiveness of the 2 MRSA interventions in preventing recurrence was important, it was also important to determine what outcomes patients and families most desired. Thus, the initial step in our study design was to engage patients and families to uncover outcomes of importance when it comes to MRSA decolonization. This study represents what appears to be the first attempt to engage patients with MRSA SSTIs in study design and tailor the measures to fit with patient-centered outcomes. Through patient and family engagement in this study, we were able to identify 9 major themes along which we could craft methods of assessment as shown in Table 2.

Table 2. Patient-centered outcome themes and associated measures.

Key patient-centered outcome themes	Trial assessment measure
Fewer MRSA[a] outbreaks	• SSTIs[b] recurrence by parental report at 6 weeks, 6 months, and 12 months • Repeat surgical (incision and drainage) procedure by parental report at 6 weeks, 6 months and 12 months
Decreased physical pain and discomfort	• Participant's level of pain and discomfort at incision site via NRS[c] pain scale at enrollment and 6 weeks
Improved emotional health, improved self-perception, decreased social stigma, increased amount of free time, and increased control over free time	• Participants' quality of life measured by parent-proxy report (or youth tool) of the PedsQL[d] 4.0 at recruitment, 6 weeks, 6 months, and 12 months • Estimated weekly time to adhere to intervention by parental report at 6 weeks
Fewer days of school or work missed and decreased financial burden	• Participants' school attendance by parental report at 6 weeks, 6 months, and 12 months • Assessment of parents' work attendance by self-report at 6 weeks, 6 months, and 12 months

[a] MRSA: methicillin-resistant *Staphylococcus aureus*.

[b] SSTIs: skin and soft tissue infections.

[c] NRS: numeric rating scale.

[d] PedsQL: Pediatric Quality of Life Inventory.

A number of these themes are interrelated, and thus the methods of assessment overlap.

The number of recurrent MRSA outbreaks was the most common theme of importance to patient advisors and parent advisors. This also happened to be the a priori primary clinically related outcome planned by the investigators. With rates of MRSA recurrence of over 70% [11-16] in some cases, it makes sense from a clinical- and patient-centered perspective that this outcome would be of paramount importance to assess in our study. Thus, we defined our primary outcome of interest as the proportion of participants with recurrent SSTIs by parental report. This would first be assessed at 6-weeks postintervention. This is a standard timeframe for assessment in emergency medicine practice and would also provide an early opportunity to check-in on study participant protocol compliance. This outcome was also assessed at 6 and 12 months. Additionally, after engaging with the participants, we believed that although avoiding infection recurrence was ideal, the avoidance of a repeat surgical procedure seemed to touch many of the key themes of importance. Thus, we also defined a secondary outcome as the proportion of participants requiring repeat I&D procedures by parental report at 6 weeks, 6 months, and 12 months. The desire for decreased pain and physical discomfort was also a major theme extracted from our interaction. Although these are partially assessed via the repeated infection/surgical intervention outcomes described earlier, we decided to assess the level of pain and discomfort at the original surgical incision site via a patient/parent report using the numeric rating scale [31] at enrollment and again at 6 weeks.

The quality of life of the patient appeared to be at the heart of the next 5 extracted themes: (1) improved emotional health, (2) improved self-perception, (3) decreased social stigma, (4) increased amount of free time, and (5) increased control over free time. Regarding improved self-perception, the literature on self-esteem identifies the self-concept of appearance as the most influential aspect affecting overall self-esteem [32]. In addition, pediatric skin diseases, particularly acquired, visible diseases such as acne or hidradenitis suppurativa, have a high negative impact on school-age and adolescent self-esteem, partly due to poor self-concept of appearance and social stigma. These diseases are associated with increased depression and suicidal ideation among older children and teens [33]. Although data on the impact of MRSA on these outcomes are lacking, we suspect that there are similarities to other skin diseases with similar features (eg, acquired and visible). How best to assess these outcomes among a pediatric population ranging in age from 3 months to 18 years of age was debated between the investigators. Ultimately, it was decided that these themes could not reliably be assessed via custom items given the age range of the participants and that an accepted and validated tool would be needed. Thus, the Pediatric Quality of Life Inventory was chosen as it is a health-related quality of life measurement tool for both healthy children and those with acute and chronic health issues [34]. This validated multidimensional tool features both self-report (5 to 18 years of age) and parent proxy-report (2 to 18 years of age) and assesses physical, emotional, social, and school functioning through 3 scores: a total score, a physical health summary score, and a psychosocial health summary score. Additionally, we felt that assessment of the time required to adhere to the study interventions would be an important measure of the burden on patients and their families.

The final themes extracted from our engagement were the desire for fewer days of school and work missed as well as a decreased financial burden. Similar to quality of life, these have not been assessed in the context of outpatient decolonization protocols. The literature on the economic costs of MRSA and other skin infections outside of direct health care system costs is limited. However, a study of the direct and indirect costs of surgical site infections (including MRSA) in adults in Spain found that 90% of surgical site infection costs were indirect costs outside of the health care system, such as lost productivity from missed work days or time spent by a family member attending to the patient [35]. Days of school and work missed secondary to dealing with SSTIs are straightforward and easy to collect via self-report. Other elements of financial burden, however, are more difficult to assess directly given the expected variability in cost/charges for medical care, insurance status, baseline socioeconomic status, and more granular issues such as the lack of a home washer and dryer requiring laundromat visits. However, it was felt that the days of school/work missed was an appropriate surrogate marker for financial burden that was feasible for the study to assess. In many cases, keeping a child home from school for illness or the circumstances surrounding a hospitalization requires at least one parent to take time away from their job to attend to the child. This could mean lost wages or arranging for childcare and potentially incurring unexpected costs.

MRSA patient-centered outcomes, to our knowledge, have yet to be explored. The literature related to quality of life or patient-centered outcomes for other skin conditions show that skin conditions do have an effect on quality of life in ways similar to our findings. For example, in 1 study, adults with atopic dermatitis reported that their condition causes avoidance of social interactions and impacts their activities [36]. A literature review related to the psychosocial effects of various chronic skin conditions found that psoriasis, a condition that causes red, scaly, painful patches of skin, impacts work, relationships, and social activities and causes anxiety and depression not only for the patient but also for cohabitants [37]. MRSA infection causes symptoms that are similar to those caused by other skin conditions, such as painful and visible skin lesions, but it also has unique aspects, particularly related to the burden of treatments such as decolonization. Thus, MRSA-specific outcomes should be measured.

Follow-Up

In total, 5 parent advisors participated in a small follow-up survey that asked them to check which of the patient-centered outcomes as described earlier were important to them when it came to MRSA treatment. In all, 3 of the 5 advisors indicated that all of the patient-centered outcomes were important to them. Items that did not receive unanimous endorsement were *improved emotional health*, *improved self-perception*, *decreased social stigma*, *increased amount of free time*, and *decreased financial burden*. This small follow-up suggests that the patient-centered outcomes uncovered during this study are promising, but additional work will be required to validate them in a larger population.

Limitations

Although these outcomes were codeveloped with a small sample size of patients and parents, all of the patients and parents had experience with skin infections such as MRSA, and a few had previously utilized or were currently utilizing bleach bath protocols. In addition, parents with children ranging from 15 months to 18 years of age and adolescents ranging from 10 to 18 years of age attended the workshop. This allowed for a range of perspectives based on patient age. HCD participatory methods also help to mitigate small numbers by getting to deeper information more quickly and allowing for discussion from the group that builds on the ideas brought up by individual members.

HCD has the same limitations as other qualitative research approaches, such as findings not being statistically representative or generalizable and potential for researcher bias in data collection and analysis. The first limitation is inherent in qualitative research and is why we see our patient-centered outcomes as a starting point that must be validated with future quantitative studies. The second was mitigated as much as possible by carefully considering the questions to be asked during our workshop and by completing analysis as a team, encouraging discussion and debate as pieces of data were grouped and theme descriptions determined.

One important limitation is that, due to limited staff resources, adolescents stayed in the room with their parents for the duration of the workshop. If adolescents had been separated from the adults, they might have been more vocal and brought up additional issues they may not have been comfortable discussing in front of their parents or other adults.

In addition, due to challenges in recruiting and retaining participants for the MEDiC trial, we were unable to effectively validate our findings or our measures or to fully assess these outcomes in our study population as planned.

Conclusions

This study represents the first attempt to engage patients with MRSA SSTIs in study design and trial development using HCD to engage patients and their families with lived experiences to determine meaningful patient-centered outcomes. Through this crucial participation, we were able to identify 9 major recurrent themes. These themes were used to develop the primary and secondary outcome measures for MEDiC, a prospectively enrolling comparative effectiveness trial launched in February 2016. The authors do not see this work as a final product, but rather a first step in inspiring the creation of a comprehensive set of MRSA patient-centered outcomes that can be measured alongside clinical outcomes in future work.

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Conflicts of Interest

PM has received research funding from Trevena Inc, the Emergency Medicine Foundation, and Society of Academic Emergency Medicine Foundation.

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Abbreviations

HCD: human-centered design
I&D: incision and drainage
MEDiC: MRSA Eradication and Decolonization in Children
MRSA: methicillin-resistant Staphylococcus aureus
SSTI: skin and soft tissue infection

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Original Paper

Health Care Consumer Shopping Behaviors and Sentiment: Qualitative Study

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Abstract

Background: Although some health care market reforms seek to better engage consumers in purchasing health care services, health consumer behavior remains poorly understood.

Objective: This study aimed to characterize the behaviors and sentiment of consumers who attempt to shop for health care services.

Methods: We used a semistructured interview guide based on grounded theory and standard qualitative research methods to examine components of a typical shopping process in a sample size of 54 insured adults. All interviews were systematically coded to capture consumer behaviors, barriers to shopping behavior, and sentiments associated with these experiences.

Results: Participants most commonly described determining and evaluating options, seeking value, and assessing or evaluating value. In total, 83% (45/54) of participants described engaging in negotiations regarding health care purchasing. The degree of positive sentiment expressed in the interview was positively correlated with identifying and determining the health plan, provider, or treatment options; making the decision to purchase; and evaluating the decision to purchase. Conversely, negative sentiment was correlated with seeking value and making the decision to buy.

Conclusions: Consumer shopping behaviors are prevalent in health care purchasing and can be mapped to established consumer behavior models.

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KEYWORDS

decision making; choice behavior; mental processes; behavioral economics; health costs; health care costs; treatment costs; cost sharing; health expenditures; out-of-pocket costs

Introduction

To mitigate rising health costs [1], employers and health insurers have increased consumer cost sharing in health insurance plan design [2,3]. Such consumer-driven health care, where consumers shoulder a greater portion of health expenditures, aims to drive down health spending by discouraging unnecessary utilization and encouraging comparison shopping for the best value care [4,5]. Despite shifting incentives [6] and related cost savings [7,8], not all savings reflect the avoidance of unnecessary or higher-priced care [9]. Recent studies have shown that simply shifting costs to consumers does not yield expected shopping behaviors. Rather, consumers often avoid necessary or preventive care [10,11]. Studies also show that consumers rarely compare prices even when tools are available [12-14], suggesting that consumer-driven health care—commonly defined narrowly by the presence of high deductibles—does not promote health care shopping.

The increased focus on consumer experience has led to investigations of consumer sentiment regarding health care encounters [15-17]. These studies highlight the applicability of automated techniques for coarsely analyzing consumer sentiment in health care and make such an analysis of large-scale unstructured data possible. However, such studies do not evaluate the association between such a sentiment and specific aspects of health care decision-making or shopping processes.

The obstacles to shopping for health care coverage and services are well documented. Information asymmetry, complexity, and patient-provider power dynamics are just a few barriers to consumer shopping [18-20]. The lack of transparency of prices—a particularly concrete and potentially addressable obstacle facing consumers—has scarcely improved in recent years despite legislative requirements and concerted efforts [21-23]. Beyond the availability and adoption of price comparison tools—and a relatively narrow definition of shopping as simply comparing prices—little is understood about how consumers could be more effectively engaged in health care shopping.

Numerous models of consumer purchasing behavior exist [24,25], but less is known about how these models apply in health care contexts. Evidence of consumer interest in engaging in health care purchasing [26] suggests opportunities for consumer-driven health care to fulfill its promise, yet also highlights gaps between consumer intention and behavior. To identify these opportunities and better explain these gaps, we sought to understand individual health care purchasing processes through a consumer lens. A deeper understanding of consumers' health care purchasing experiences would enable health care organizations and policy makers to design interventions to efficiently engage consumers and help improve consumer value in the US health care market. Specifically, understanding the aspects of shopping that consumers find particularly challenging, or gratifying, should aid in the development of interventions to facilitate such processes.

Methods

Overview

We recruited a convenience sample (N=54) of individuals aged 18 to 98 years based on a study protocol and obtained a consent form approved by the human subjects review committee of the Harvard Kennedy School. To preserve participant confidentiality, considering reidentifiability, consent forms do not provide for release of individual data. Intensive interviews [26] were conducted by phone or in person by the senior investigator, an experienced qualitative interviewer, and 2 Masters-level student researchers trained by the senior investigator. All but 5 interviews were recorded and professionally transcribed; where participants did not consent to recording or recording was not available, researchers captured participant responses in detailed interview notes.

Interviewers categorized each participant by insurance type, age, and gender based on self-reports. Similarly, participants were categorized by health-related characteristics including health status and utilization, either explicitly articulated by participants or inferred by the researchers. In cases where the participant was not explicit and it was not clear from context, we categorized participants as "not reported" for that measure.

Interviewers used a semistructured interview guide developed by the research team, and based on grounded theory, a systematic empirical research methodology was used to construct the theory inductively via methodical data gathering and analysis [27,28]. Interviews lasted approximately one hour and were organized around components of a typical shopping process, such as the consumer decision process depicted in Blackwell/Miniard's model (Figure 1) [25], adapted to include the following:

1. Identifying the need or desire for a health care purchase (need recognition)
2. Determining and evaluating options to meet that need or desire (search/prepurchase evaluation of alternatives)
3. Making the decision to purchase (purchase/consumption)
4. Evaluating the decision to purchase (postconsumption evaluation/divestment)

Within this shopping framework, we examined behaviors relevant to consumer value capture, such as trying to understand costs before seeking care or negotiating the cost of care before or after a service, advocating for one's self, or making trade-offs such as paying more for convenience or accepting low-quality service to save money.

We also sought to identify barriers to traditional consumer behaviors and value capture. Barriers were either systemic (eg, administrative hassles or lack of price transparency) or consumer limitations that constrained their ability to capture value (eg, ignorance or confusion about how to capture value).

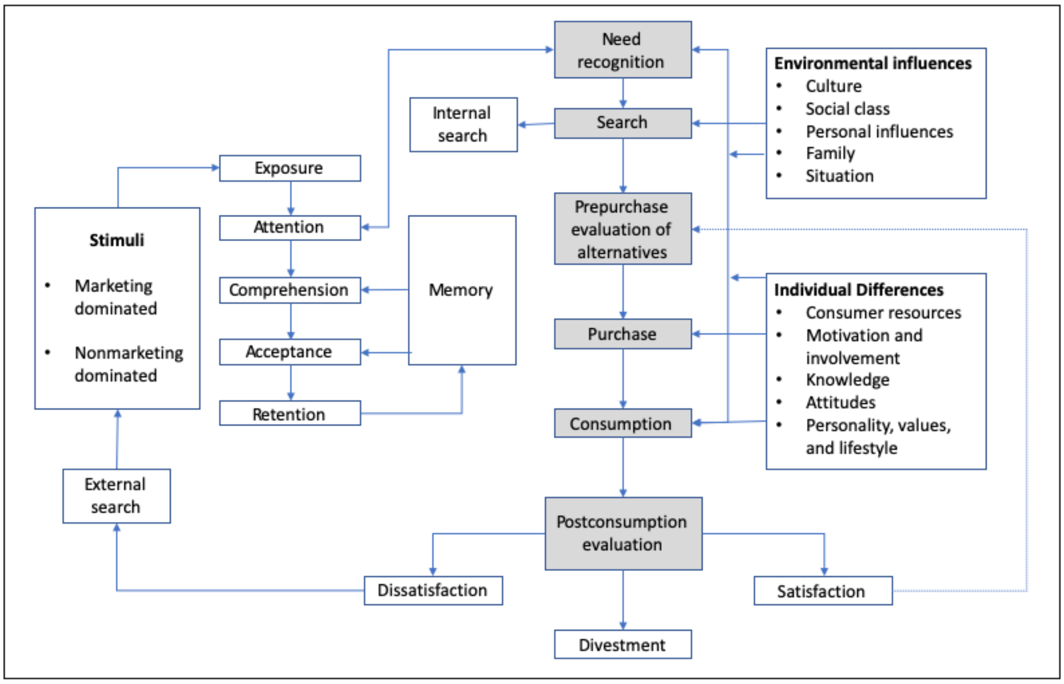
Finally, we sought to capture participant sentiment. Positive sentiments included feelings such as gratitude, relief, peace of mind, or optimism. Negative sentiments included anger, frustration, despair, anxiety, or pessimism.

Data Analysis

Transcripts and interview notes were processed using Dedoose (SocioCultural Research Consultants, LLC), an application for managing, analyzing, and presenting qualitative and mixed methods research data [29]. The authors developed an initial code set organized around the typical shopping process components investigated in the interview guide. Following grounded theory methods [26], additional codes were created to capture emergent themes such as sentiments about shopping and beliefs about the health care system. Researchers coded each transcript, and the senior investigator reviewed all codes in all transcripts to ensure consistency in the application of codes.

For the examination of sentiment associated with behavior, specific codes were identified to indicate either positive or negative feelings. Positive sentiment included the codes for optimism bias, peace of mind/comfort, and gratitude/relief. Negative sentiment included the codes for pessimism bias, vulnerability, anger/frustration, despair/desperation, fear/anxiety, and financial anxiety/concerns about cost.

Figure 1. Consumer behavior model.



As this was intended primarily as a hypothesis-generating study, the majority of analyses were descriptive, examining the frequency with which participants described engaging in particular behaviors or encountering particular scenarios. We calculated power to detect a given theme as described by Fugard and Potts [30]; for a theme with 10% population prevalence, power exceeds 90% to detect that theme at least three times in an interview cohort of this size.

The authors also examined the extent to which expressions of positive or negative consumer sentiment were associated with the discussion of each shopping process stage. For these tests, Pearson correlations were utilized to examine the relationship between the frequency of code pairs, with a sensitivity analysis using linear regression to adjust for effects of age and gender. All analyses utilized Stata SE 13.1 [31]; figures were generated using R version 3.5.0 [32]. While nominal *P* values are reported, for secondary analyses, we focused on the magnitude of effect (eg, correlation).

Results

Participant Details

A total of 54 interviews were completed with individual consumers. All participants were currently insured; 65% (35/54) were women, and the mean age was 43 (SD 16.23) years (Table 1). Despite the majority (49/54, 91%) reporting being in good-to-excellent health, health care utilization was estimated as moderate or high for 80% (43/50) of participants. More than half the participants (30/54, 56%) reported either a chronic condition or a past catastrophic accident or emergency requiring follow-up care. A minority (5/54, 9% of all participants) reported a current or previous cancer diagnosis.

Participant responses revealed that in addition to 4 basic stages in a shopping process, seeking value before a purchase and assessing value following a purchase were important components in the health care purchase processes. Thus, the analysis incorporated these 2 concepts:

- Stage 1: Identifying the need or desire for a purchase in a health care context
- Stage 2: Determining and evaluating options to meet that need or desire
- Stage 3: Seeking value (a subset of determining/evaluating options)
- Stage 4: Making the purchase decision
- Stage 5: Evaluating the purchase decision in terms of quality and/or satisfaction
- Stage 6: Assessing value (a subset of evaluating the purchase decision)

The distribution of participant responses by shopping process stage is reported in Multimedia Appendix 1. Nearly every participant addressed each shopping stage at least once; the shopping stage discussed most often was Stage 2 (determining and evaluating options), followed by Stage 3 (seeking value), and Stage 6 (assessing value). Participants with individual insurance (ie, purchased on a state or federal health insurance marketplace) more frequently mentioned Stage 2 (determining and evaluating options; t_{52}=2.90; P=.007).

We also categorized the most commonly cited consumer shopping behaviors, summarized in Table 2; intensity, measured by the average number of mentions per participant, is depicted in Table 3. Almost all participants (53/54, 98%) had experience paying out-of-pocket or sharing in health care or coverage costs; these experiences were discussed more than 5 times per interview, on average.

Table 1. Participant characteristics (N=54).

Characteristics	Values
Age (years), mean (SD)	43.44 (16.23)
Gender, n (%)	
Female	35 (65)
Male	18 (33)
Intersex	1 (2)
Insurance type, n (%)	
Employer-sponsored	24 (44)
Individual	12 (22)
Medicare	5 (9)
Other insurance[a]	13 (24)
Health status, n (%)	
Excellent	12 (22)
OK/pretty good	37 (69)
Not great	3 (6)
Not reported	2 (4)
Utilization, n (%)	
Low	11 (33)
Moderate	26 (2)
High	35 (65)
Presence of a chronic condition or prior accident/emergency, n (%)	
Yes	30 (56)
No	14 (26)
Not reported	10 (19)

[a] Other insurance includes student insurance or Medicaid.

Most participants (53/54, 98%) discussed seeking or comparing price information or going to a provider where such information was explicit. Seeking cost information before a service was reported by 72% (39/54) of participants and comparing provider prices was reported by 56% (30/54) of participants.

All participants reported seeking value in some way, by responding to financial incentives, using health savings vehicles, or pursuing workarounds to capture the economic value. Seeking value had the highest number of mentions on average (6.48 per interview). Most participants (45/54, 83%) described some form of negotiating—one type of value-seeking behavior—which could include bargaining with a provider (23/54, 43%), arguing about a medical bill after a service (17/54, 31%), or negotiating with an insurance company for the approval of requested coverage (17/54, 31%). Participants who discussed negotiating with a provider referenced dentists (15/54, 27%), psychotherapists (10/54, 18%), out-of-network providers not covered by their insurance plans (10/54, 18%), hospitals or other providers with whom the participant had an outstanding balance (8/54, 15%), and other types of providers (24/54, 45%).

Though cost was a consideration for 85% (46/54) of participants, two-thirds (36/54, 67%) also discussed situations in which they were price insensitive or where factors other than price drove their care or coverage decisions. Significant positive correlation between price insensitivity in provider selection and price insensitivity in health plan selection was observed (r=0.32; P<.02). An adjustment for participant age and gender in regression models did not meaningfully change this association. Individual insurance was associated with discussing factoring costs (t_{52}=3.30; P=.002), whereas employer-sponsored insurance was associated with price insensitivity (t_{52}=−2.8; P=.008). Brand was not a dominant factor in participant selection of provider or health plan, arising in just 22% of interviews.

The majority of participants had experienced systemic barriers—billing errors or insurance policies blocking needed services (Table 4)—to capturing value. All participants expressed personal barriers such as their own ignorance or attitude. Notably, 91% (49/54) of participants articulated lack of trust—reflected in questioning their provider's authority or the motivations of their provider or health plan. Table 4 includes the frequency of each type of barrier cited.

Table 2. Frequency of explicit consumer shopping behaviors.

Consumer behavior	Participants (N=54), n (%)	Mentions, mean (SD)
Paying for care	53 (98)	5.07 (3.53)
Seeking/comparing/knowing prices	53 (98)	5.22 (3.88)
Seeking cost/price before getting care	39 (72)	1.78 (2.00)
Comparing prices/shopping around for better price	30 (56)	1.17 (1.49)
Knowing costs/seeing provider with flat fees	21 (39)	0.62 (1.13)
Negotiating/arguing bills	45 (83)	3.26 (3.06)
Negotiating with a provider[a]	23 (43)	0.98 (1.58)
Arguing a bill after a service	17 (31)	0.67 (1.67)
Negotiating with the insurance company for approval of a requested service or drug	17 (31)	0.80 (1.38)
Exhibiting self-advocacy/empowerment	48 (89)	4.52 (3.76)
Seeking value (eg, responding to incentives, using health savings)	54 (100)	6.48 (4.39)
Making trade-offs in decision making	46 (85)	2.94 (2.37)
Factoring brand in provider or plan selection	12 (22)	0.44 (1.02)
Factoring cost in plan/provider/treatment selection	46 (85)	4.26 (3.86)
Factoring cost in health plan selection	28 (52)	1.22 (1.60)
Factoring cost in provider selection	15 (28)	0.43 (0.81)
Factoring cost in treatment decision	37 (69)	2.61 (3.37)
Price insensitivity	36 (67)	1.72 (1.78)

[a] Of the participants who discussed negotiating with providers, 27% (6/23) discussed negotiating with dentists, 18% (4/23) with psychotherapists, 18% (4/23) with out-of-network providers, 14% (3/23) with hospitals or other providers with whom the participant had an outstanding balance, 9% (2/23) discussed negotiating for medications or with pharmacies, and 5% (1/23) each with an optometrist, with a chiropractor, in regular doctor visits, and in medical tests.

Table 3. The intensity of discussion of consumer shopping behaviors.

Consumer behavior	Participants, n (%)	Mentions, mean (SD)
Paying for care	53 (98)	5.07 (3.53)
Seeking/comparing/knowing prices	53 (98)	5.22 (3.88)
Negotiating/arguing bills	45 (83)	3.33 (3.08)
Exhibiting self-advocacy/empowerment	48 (89)	4.52 (3.76)
Seeking value (eg, responding to incentives, using health savings)	54 (100)	6.48 (4.39)
Making trade-offs in decision-making	46 (85)	2.94 (2.37)
Factoring brand in provider or plan selection	12 (22)	0.44 (1.02)
Factoring cost in provider/plan/treatment selection	46 (85)	4.26 (3.86)
Price insensitivity	36 (67)	1.72 (1.78)

Table 4. Barriers to consumer shopping behavior.

Barrier type	Participants (N=54), n (%)	Mentions, mean (SD)
Systemic barriers	46 (85)	4.52 (3.9)
Personal barriers	54 (100)	10.5 (6.59)
Attitudes (eg, denial, resignation)	40 (74)	1.74 (1.67)
Confusion/ignorance	46 (85)	4.44 (4.33)
Lack of trust	49 (91)	4.31 (3.06)

Association Between Extent of Positive or Negative Sentiment

The association between extent of positive or negative sentiment expressed by participants and the extent to which each shopping stage was discussed was also examined. Positive sentiment was significantly and positively correlated with Stage 2 in the shopping process (identifying and determining health plan, provider, or treatment options; *r*=0.58; *P*<.001), Stage 4 (making the purchase decision; *r*=0.45; *P*<.001), and Stage 5 (evaluating the purchase decision; *r*=0.38; *P*=.004). Negative sentiment was significantly and positively correlated with Stage 3 (seeking value; *r*=0.30; *P*=.02) and Stage 4 (making the purchase decision; *r*=0.31; *P*=.006).

Finally, we examined the association between positive or negative sentiment and the extent to which each consumer shopping behavior was reported. Positive sentiment was not statistically significantly correlated with any of the behaviors. Negative sentiment was significantly and positively associated with paying for care out-of-pocket or cost sharing (*r*=0.40; *P*<.03), negotiating (*r*=0.45; *P*<.001), and self-advocacy (*r*=0.42; *P*=.001).

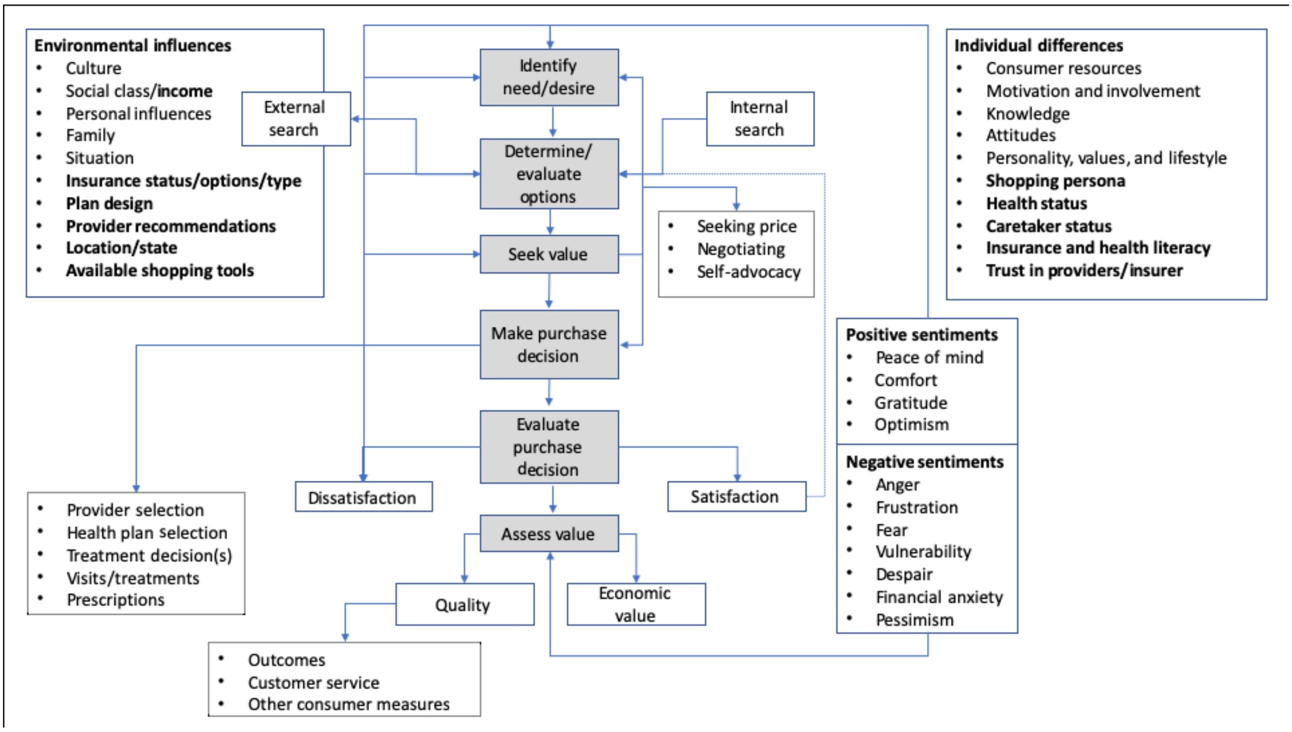
Discussion

Principal Findings

In this investigation of health consumerism among 54 insured individuals across a range of ages with generally high health care utilization, explicit consumer shopping behaviors—even those not typically associated with health care decision making—were prevalent, though not always successful. Participants perceived pervasive barriers to engaging in health care shopping. Most participants experienced systemic or administrative barriers, and all exhibited personal barriers related to their attitude, knowledge, or trust in the system.

Despite barriers, these results indicate that health care purchasing processes, as different as they may appear from other purchases, could be mapped to established consumer behavior models. We have offered an adaptation of Blackwell/Miniard’s model to account for the findings on the specific nature of and feelings about health care shopping processes [25]. Figure 2 visually depicts findings of the most salient steps and factors in health care purchasing. This model provides a potential alternative to prevailing assumptions that health care purchasing does not reflect traditional consumer shopping behaviors; it also invites further refinement to establish a standard framework for health care shopping processes.

Figure 2. Consumer health care shopping process.



Participants who had purchased individual insurance on either the state or federal health insurance marketplace discussed Stage 2 of the shopping process (determining and evaluating options) more than those with employer-sponsored insurance, suggesting the possible influence of the Affordable Care Act on consumer orientation toward shopping for health insurance.

We found that 98% (53/54) of participants had engaged—or tried to engage—in behaviors relating to seeking, comparing, or knowing prices of care. Specifically, in our study, 72% (39/54) of participants reported seeking information about the cost of care, and 50% (27/54) reported comparing prices or looking for a lower price via an alternate provider. Our study participants had greater prevalence of these behaviors than other studies; Public Agenda [26] found that 50% of participants surveyed had tried to find price information and 20% tried to compare prices between providers. Mehrotra et al [13] found that 13% of subjects had tried to find price information and 3%

had tried to compare prices. Both Public Agenda (n=2062) [26] and Mehrotra et al (n=1904) [13] used structured survey instruments with large-scale samples. This study's smaller sample may be biased toward people with more health care experiences than a general population, and intensive interviews may be more sensitive than survey instruments. Additionally, the open-ended nature of our study may reflect broader interpretation of these behaviors, compared with a close-ended survey question. These results support others' findings that there is widespread consumer interest in knowing the prices of health care services.

Brand, which is central in many shopping processes and is the focus of billions of health care marketing dollars each year [33], was not a dominant factor in participant provider or health plan selection, with less than a quarter of participants reporting it. This finding may reflect opportunities for more effective branding efforts, or it may reflect the need for health plans and providers to focus on other measures, such as affordability. Finally, it may simply reflect consumers' own underreporting of the importance of brand as a factor in their selections.

Cost was a consideration for 85% of participants, despite 44% of participants having employer-sponsored insurance, a group with generally lower financial barriers to accessing health care services [33]. This result likely reflects the impact of increased use of high-deductible plans among employers.

The authors also sought to understand how participants feel about health care purchasing by examining how the amount of discussion of a particular shopping stage relates to the amount of positive or negative sentiment expressed by participants. Positive sentiment was significantly and positively correlated with identifying options, making purchase decisions, and evaluating purchase decisions. One previously uninsured participant positively evaluated his individual insurance purchase: “I'm very comfortable with it…I'm pretty pleased to have it.”

Negative sentiment was significantly and positively correlated with seeking value and making the purchase decision. One participant found seeking value following an unsuccessful surgery infuriating:

> *I have to get revision surgery. If I was not happy with another service, I wouldn't pay the bill. I would fight the bills. In this case, I have scar tissue that is causing me problems, I still can't breathe. Why am I still on the hook for a little bit of money?*

Positive sentiment associated with identifying options, making decisions, and evaluating decisions may suggest participants' appreciation for the availability of options and the opportunity to make decisions. Negative sentiment associated with seeking value likely reflects the frustration consumers expressed over systemic barriers to finding cost information and, more generally, to the high cost of health care coverage and services. Negative sentiment associated with making purchase decisions may suggest discomfort among some consumers with available options or a general unease or distaste for needing to function as a health care decision maker.

It cannot be concluded, based on this study design, whether these associations are causal or reflective of more complex relationships. However, these correlations suggest the possibility of interesting relationships between sentiment and consumer shopping processes in health care, which merit further investigation to clarify the nature of the relationships.

The authors also sought to understand how sentiment related to engaging in explicit consumer shopping behaviors and found no relationship between consumer shopping behaviors and positive sentiment. However, negative sentiment was significantly associated with paying out-of-pocket or cost sharing, negotiating, and self-advocacy. As noted, the design of this study does not allow for the determination of causality, but the interviews suggest the relationship may be bidirectional. For example, after a procedure, one participant tried unsuccessfully to negotiate with a doctor who had billed insurance for two separate procedures:

> *[The doctor] didn't care, since this is the way they bill it…they expect their money. I paid for that other part, which I didn't think I should have paid for, and I told them, I said, “You've just lost yourself a patient and other references.“*

Conversely, another participant's negative experience led to enhanced consumer behaviors. Undergoing cancer treatment, she experienced a lack of personalization and inadequate access to her providers. These negative experiences led her to more active self-advocacy:

> *I'm my own advocate. My husband's my advocate. We are the quarterbacks. . . we learned we had to play [that role]. I did not assume I would need to do this. . . this was my first experience with health care where I realized it's not up to them, it's up to me. . .*

Further investigation could illuminate the nature of these relationships and the prevalence in a general population.

Limitations

Multiple limitations in this study should also be noted. First, as a convenience sample, selection bias cannot be excluded in the sample; those who agreed to participate may be more likely to have health care experiences to discuss. Second, the grounded theory method does not search for objective “truth” but rather develops theories based on empirical qualitative data [27,28]. As such, it does not deduce testable hypotheses from existing theories. Critics find grounded theory specifically overly reliant on empirical data, and qualitative methods generally to be anecdotal or impressionistic. However, proponents point to the power of qualitative methods to provide a conceptual understanding of studied phenomena and emergent, original theories [27]. Additionally, temporal, spatial, geographic, and personality or psychological traits or propensity toward positive or negative sentiments may influence participant responses; other than noting optimism or pessimism biases and including those in the sentiment analysis, these factors are not considered [34,35].

Finally, the limited sample size, and particularly small numbers of some subgroups within the sample, may impact the generalizability of our results and preclude additional hypothesis testing.

Nonetheless, these results help to illuminate consumers' experiences with and attitudes toward health care purchasing. Further examination of the differences by demographic segment and type of purchase (eg, care vs coverage) will advance this effort and determine if these findings apply to different populations. Additionally, large-scale surveys would confirm or refine these findings.

Conclusions

More generally, these results confirm widely reported obstacles to consumer shopping behaviors in health care, from structural barriers like lack of price transparency to individual constraints like information asymmetry or confusion. On the contrary, these results also reinforce the potential role of market forces in health care and the conceptual relevance of consumer shopping behavior frameworks. Similarly, narrow definitions of consumer-driven health care—as simply high-deductible health plans—ought to be broadened to include a wider range of behaviors and incentives. Such reframing would enable future studies to explore the discordance between consumers' desire to engage and their ability to do so to capture value in health care purchasing. Recognizing that consumers do shop for health care, and understanding how they shop for health care, are crucial steps in designing interventions to enhance this process.

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The authors confirm that each author participated sufficiently in the work to take public responsibility for the content. Individual authors contributed to (1) the conception and design or analysis and interpretation of data and to (2) drafting the study or revising it critically for important intellectual content. All authors granted final approval of the version to be published. No external funding was received for this study.

Conflicts of Interest

RP holds equity in Psy Therapeutics and Outermost Therapeutics and has served on advisory boards or provided consulting to Genomind, Psy Therapeutics, RIDVentures, and Takeda. The other authors have no conflicts of interest to report.

Multimedia Appendix 1
Frequency of shopping stages.
[DOCX File , 14 KB - jopm_v12i2e13924_app1.docx]

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