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Review

A Snapshot of Community Engagement in Research in the Context of an Evolving Public Health Paradigm: Review

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Abstract

Background: Community engagement is a work in process. Researchers continue to refine the process of collaboration and look to best practice and lessons learned for guidance in this relatively new model.

Objective: The aim of this study was to provide a snapshot of whether community engagement has been included in the design and implementation of research initiatives in Australia, Canada and the United Kingdom. The secondary aim is to identify the underlying themes present, to identify theories and tools that drive research.

Methods: A literature search was performed to identify studies that have been conducted to reduce the weight of the general population.

Results: The results of the study, which focused on the field of weight loss, indicate that scientific and technological advancements are the primary drivers of research. However, these new research initiatives have largely been undertaken in the absence of community engagement.

Conclusions: The study concludes that initiatives need to adapt to a wider range of stakeholders, develop equitable community engagement platforms and take into consideration.

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KEYWORDS

Community engagement, weight loss, paradigm shift, public health policy

Introduction

Early phases of public health focused on creating physical infrastructure, improving sanitary conditions, and fighting and containing known infectious diseases [1]. This model addressed the immediate needs of the population and set the fundamental basis for modern public health systems. Further movements, particularly towards the end of the 20th century, addressed the role of individual behavior on noncommunicable diseases and premature death and focused on disease prevention [2]. A modern public health emerged in the 1990s with a consensus in the international community that health promotion guided by the Ottawa Charter principles constituted public health [1]. The significance of the new public health was that it recognized health as a key determinant of the quality of life of individuals and specific populations. It incorporated elements from previous

models to create a new movement with a more unified, community-based and interconnected path between the many components of public health [3,4].

Modern public health continues to evolve and is responsive to globalization, and political and physical environments. As with early phases of public health, modern public health still places importance on physical infrastructure; however, the aim is to enhance its value and effectiveness with the addition of social support and acknowledgement of behavioral factors; creating a more holistic, intersectorial approach to health issues [5]. The beginning of this century has seen a further extension of modern public health where factors that are not traditionally health related, such as environmental sustainability and intellectual property, are also taken into consideration when reacting to health issues and developing initiatives [6].

While there have been shifts towards more social movements to improve the health of communities, they are still primarily expert driven, top-down initiatives [4]. Community engagement is a work in process. Health professionals and researchers continue to review and refine this process of collaboration and look to best practice and lessons learned for guidance in this relatively new model [7,8]. The aim of this study was to provide a snapshot of whether community engagement has been included in the design and implementation of research initiatives in Australia, Canada and the United Kingdom. The secondary aim is to identify the underlying themes present, to identify theories and tools that drive research.

For the purpose of this study, the field of nutrition, specifically initiatives that aim to support weight loss in a general population, will be investigated. The field of weight loss was selected as there is a growing, worldwide effort to address the impact of the increasing incidence, mortality and cost to society of overweight and obese populations. It was also selected as eating is an everyday activity and it can be reasonably expected that communities are involved in research within this field. Furthermore, while the outcomes of weight loss interventions have been reviewed, [9-16] there is little evidence on how communities have been engaged in research and the trends driving new research initiatives.

Methods

The lead researcher performed a literature search to identify studies that have been conducted to reduce the weight of the general population. The study covered a number of key areas: public health, nutrition, health promotion, and sociology. For this reason, the lead researcher used the PubMed database to collect sources from each sector needed to achieve a balanced and comprehensive result.

The broad search included the title/abstract search terms “weight loss” and “intervention”, excluding the Medical Subject Headings “Surgical Procedures,” “Operative,” and “Drug Therapy,” with the inclusion criteria set to randomized controlled trials, studies published between 2000 and 2014, and language in English. The search restricted studies to those conducted in Australia, Canada and the United Kingdom as these comprise countries with similar public health systems.

Studies excluded were those not implemented in Australia, Canada or the United Kingdom; if the study focused on a subpopulation with a specific disease or condition; studies that included a pharmacological intervention; and studies that did not have an outcome or measurement of weight loss.

The lead researcher classified the included publications according to the focus of the intervention and grouped these into themes. This was achieved by determining the theory or element that the interventions tested within each study. The lead researcher conducted the literature search and process of classification twice to assure accuracy and consistency of classification.

The lead researcher recorded the number of studies in each theme and used this information to rank themes in an effort to

understand the drivers or factors that influence the development of research initiatives. The lead researcher ranked the theme that yielded the most studies first, and the theme that yielded the least studies last.

To understand the level of community engagement included in the studies, the lead researcher reviewed each study and recorded indicators of community engagement in relation to (1) study development (whether the research group engaged a consumer or patient group/representative in the development of the protocol before the intervention was finalized), (2) future implementation (whether consumer or patient engagement is recommended as part of next steps or future work) and (3) acknowledgement (whether the research group acknowledged the contribution of participants in the study). It is important to note that acknowledgement on its own may not function as an indicator of community engagement; however, it has been included as a gauge that may be used in future studies to measure whether there is an increase in basic acknowledgment of participants in studies.

Results

The initial search of studies between 2000 and 2014 yielded 250 publications. 164 articles did not meet the inclusion criteria and 86 publications representing 53 individual studies met the inclusion criteria. See [Figure 1](#).

Following a review of all studies, the lead researcher identified 13 classification groups and then ranked studies within each classification (see [Table 1](#)). This resulted in nine studies (17.0%) investigating “Macronutrients” and weight loss, which was the most common theme, followed by interventions that tested “Caloric restriction +/- exercise” and “Counseling/Additional Therapeutic Contact/Behavioral Therapy/Lifestyle intervention” (n=8, 15.5% respectively), “Commercial weight loss programs” (n=7, 13.2%) and “Web-based/App-based/Text messaging/Electronic Device” (n=6, 11.3%). The remainder of the themes, including “Community-based interventions”, had three or fewer studies. The number of studies listed within each of the categories ranged from nine to one (see [Table 2](#)).

The lead researcher then identified five broad classifications resulting in 22 studies (41.5%) responding to “Scientific advancements/Investigating biological interactions and weight loss;” 13 studies (24.5%) responding to “New technologies or commercial opportunities;” 10 studies (18.9%) responding to “Psychological/Behavioral Theories;” five studies (9.4%) responding to theories in “Community-based interventions;” and three studies (5.7%) responding to the need to test the “Efficacy of information or guidelines.”

In relation to documented community engagement within publications, two studies (3.7%) documented evidence of community engagement in the development of the protocol, four (7.5%) noted that they would incorporate community engagement activities in future, related initiatives, and 17 studies (32.1%) acknowledged and thanked the people that participated in the study. 35 studies (66.0%) had no documented form of community engagement across the three indicators (see [Table 3](#)).

Figure 1. Search yield for study literature search.

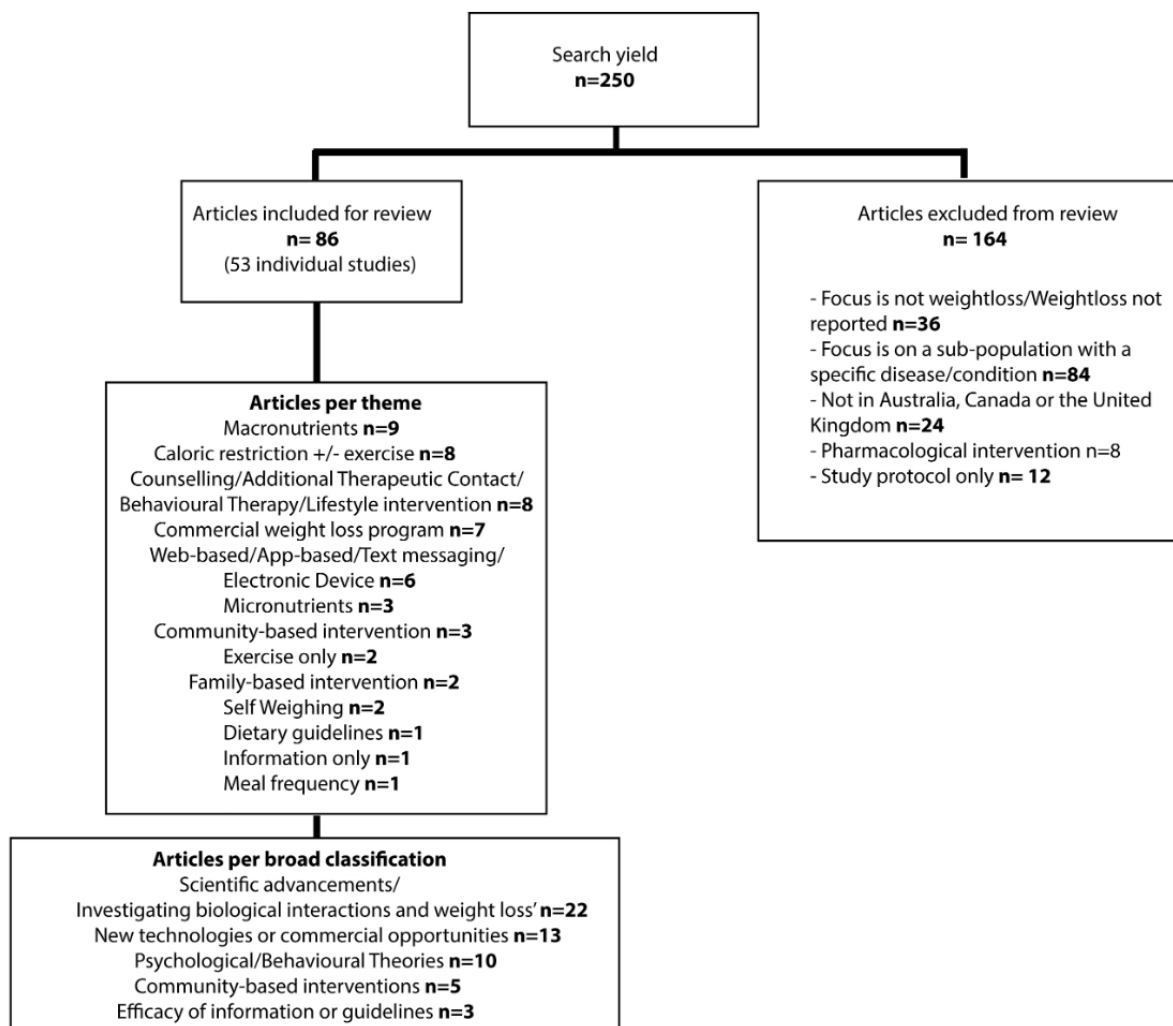


Table 1. Theme descriptions.

Themes	Definitions
Macronutrients	Studies that compare or investigate the interaction of macronutrients and weight loss
Caloric restriction +/- exercise	Studies that investigate caloric restriction in weight loss, some with and some without exercise
Micronutrients	Studies that compare or investigate the interaction of micronutrients and weight loss
Exercise only	Studies that include interventions that use exercise only to reduce weight
Commercial weight loss program	Studies that compare the efficacy of commercial weight loss programs
Web-based/app-based/text messaging/ electronic device	Studies that test a web-based platform or app-based platform or text messaging or electronic device to deliver a weight loss intervention.
Self-Weighing	Studies that test the implication of self-weighing on weight loss
Counseling/additional therapeutic contact/ behavioral therapy/lifestyle intervention	Studies that use additional therapeutic contact or behavioral therapy or lifestyle interventions as the focus of a weight loss intervention
Community-based intervention	Studies that are based in local communities and a developed based on cultural or social interactions
Family-based intervention	Studies that are based on the participation of various family members within the one intervention
Dietary guidelines	Studies that test the efficacy of published clinical guidelines on weight loss
Information only	Studies that provide participants with written information only as an intervention to support weight loss
Meal frequency	Studies that investigate the frequency of meals and the impact on weight loss

Table 2. Results by themes and broad classifications.

Themes	No of Studies				Ranking	Broad classification	Broad classification
	Australia	Canada	UK	Total: Themes			
Macronutrients	6	2	1	9	1	Scientific advancement/ investigating biological interactions and weight loss	22
Caloric restriction +/- exercise	1	7	0	8	2		
Micronutrients	1	2	0	3	6		
Exercise only	0	1	1	2	8		
Commercial weight loss program	1 ^a	5	2 ^a	7	4	New technology or commercial opportunities	13
Web-based/ app-based/ text messaging/ electronic device	4 ^a	0	3 ^a	6	5		
Self-weighing	0	0	2	2	8	Psychological/ behavioral theories	10
Counseling/ additional therapeutic contact/ behavioral therapy/lifestyle intervention	4	0	4	8	2		
Community-based intervention	2	0	1	3	6	Community-based intervention	5
Family-based intervention	1	0	1	2	8		
Dietary guidelines	1	0	0	1	11	Efficacy of information or guidelines	3
Information only	0	0	1	1	11		
Meal frequency	0	1	0	1	11		

^aNote: One study was conducted in both Australia and the United Kingdom.

Table 3. Indicators of Community Engagement (CE) within studies reviewed.

Themes	CE Development	CE Future	CE Acknowledgement
No. of Studies			
Macronutrients	0	0	2
Caloric restriction +/- exercise	0	0	2
Micronutrients	0	0	0
Exercise only	0	0	1
Commercial weight loss program	0	0	1
Web-based/ app-based/ text messaging/electronic device	0	1	3
Self-weighing	0	0	0
Counseling/additional therapeutic contact/ behavioral therapy/lifestyle intervention	0	1	4
Community-based intervention	1	1	2
Family-based intervention	0	1	1
Dietary guidelines	0	0	1
Information only	1	0	0
Meal frequency	0	0	0
Total	2	4	17

Discussion

The majority of the studies reviewed were conducted in response to “Scientific advancements/Investigating biological interactions and weight loss” and “New technologies or commercial opportunities”, that is, they were primarily advancing and testing new knowledge (such as micronutrient or macronutrient involvement in weight loss) or tools (such as the Internet and electronic devices to deliver interventions). This is to be expected and encouraged in an evidence-based health sector. What is of interest is that there were only a few studies that were community-based and very few studies that reported significant community engagement. While the subject matter for this review was interventions that aim to reduce weight and the results cannot be generalized to all public health areas, it gives us an indication that in public health research, the notion of community engagement and empowerment has not been fully leveraged.

This is important because the foundation of public health revolves around empowerment, community involvement, a multidisciplinary alliance and achieving equity in health [17]. Empowerment refers to the ability of people to acquire an understanding and control over personal, social and economic influences on their health so that they are able to act in a way that will improve their life situation [18]. These are all factors that are difficult to measure and in the context of public health it is a challenge as it may not always be possible to report community engagement and empowerment in a way that satisfies traditional impact measures.

Another challenge is that the emphasis on empowerment is often in contrast with equally influential notions of evidence-based decision making including cost-effectiveness and population health approaches. This is largely driven by stakeholders and decision makers often being more concerned with the ability to measure outcome and define empirical success rather than the value that the target population places on the initiative itself [19]. In the context of this review, it should be noted therefore that there may have been more community engagement within the studies reviewed, but it was not reported as it is not generally valued or requested in scientific literature.

It appears then that a key challenge in public health is to build high quality and appropriate standards for evidence-based evaluation that the community, researchers and policy makers can mutually benefit from.

In the public health setting, the promotion of health is defined as a process in which individuals are able to increase control over their health resulting in an improvement in their life [20]. While it is not a new document, the Ottawa Charter continues to provide public health practitioners with guidance from a combination of its five action areas. Within the five action areas there are two key elements that are particularly relevant to public health policy. The first is to “Build healthy public policy”, and the second is to “Strengthen Community Actions”. The first element relates to the regulatory aspects of public health where policies and laws are created to enforce health promotion initiatives while the second element relates to empowerment and the ability of communities and patients to set priorities,

make decisions, plan and implement programs that help them to improve their health outcomes. While these are both extremely important elements, they are potentially conflicting and it is not clear whether they can coexist in the context of modern public health, as was evidenced in this review where a number of high quality weight loss studies reported detailed clinical and policy-related outcomes. However, the vast majority of studies did not report or measure community engagement.

When we look at the results of the community engagement indicators in this review, there were only five studies that demonstrated an effort to engage patients in the development of their research protocol or future research initiatives. It is clear that this is an area that researchers have not completely embraced as part of their research process, yet patients and the general public are demanding an increased level of accountability from health professionals and policy makers regarding allocations of health resources by governments and health care providers [21-25]. This is important because research that includes collaboration between health professionals, knowledge through research, and drawing upon patients and community members’ knowledge about their health, safety and well-being are most effective, particularly when they are complemented with an analysis of the needs and expectations of the community [26-28]. Acknowledging the differences in community needs and expectations may firstly avoid the development of a problematic or inappropriate health policy or initiative [29], and furthermore can assist in creating a supportive environment and improve an individual’s ability to access all appropriate and available services and treatments [30-35].

There are, however, conflicting paradigms in health that challenge our ability to engage the community and drive research and policy that addresses individual needs. While population health approaches aim to deliver services and initiatives that serve the greater population, it is often at the risk of bypassing minority groups and potentially increasing the gap in health inequality [26,36]. Public health has progressed from a largely reactive model, to a preventative model. The next step is to make it a more proactive movement. That is not to say that it should not be reactive or preventative, but rather a combination of various elements of previous public health models. The differences between the old and the new public health are relatively subtle and are in many ways the result of the different context and environments in which public health exists [37]. Moving forward, the sector will need to acknowledge that there are many determinants of health and risk factors, some of which will be restricted to small subpopulations, which can be addressed by also using multi-sectoral and innovative partnerships to implement practical work plans [27,28].

This evolution means that public health professionals will be required to have expertise not only in health, but knowledge of a wider range of disciplines, an understanding of community engagement methods and incorporate a multidisciplinary approach to health in their decision making. Another explanation for the poor level of community engagement in this review may therefore be a lack of support and training for researchers to implement community engagement activities. This is important because those who create health policies are also now required

to take into account the varying contexts that affect health outcomes [38] and if the community view is not included in the research that informs policies, the ability to make informed decisions may be compromised. This evolution has certainly created a more complex platform for public health; however, it also provides valuable opportunities for collaboration with an extended range of stakeholders including patients and the community, who can contribute additional knowledge, experience and set the expectations of public health initiatives.

Conclusion

This review provides a demonstrative snapshot of the level of community engagement in one area of public health research.

While it is not common to all areas of public health, it is clear there are many drivers of public health initiatives and that scientific and technological advancements are the primary drivers of research. However, these new research initiatives have largely been undertaken in the absence of community engagement. Development and evaluation of research and public health initiatives need to adapt to a wider range of stakeholders including looking for best practice community engagement, embracing new prospects for collaboration, developing new and equitable patient and community engagement platforms and taking into consideration the more complex social environment as well as individual needs.

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Conflicts of Interest

None declared.

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Abbreviations

CE: community engagement

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Review

Interventions to Improve Movement and Functional Outcomes in Adult Stroke Rehabilitation: Review and Evidence Summary

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Abstract

Background: Patients who have had a stroke may not be familiar with the terminology nor have the resources to efficiently search for evidence-based rehabilitation therapies to restore movement and functional outcomes. Recognizing that a thorough systematic review on this topic is beyond the scope of this article, we conducted a rapid review evidence summary to determine the level of evidence for common rehabilitation interventions to improve movement/motor and functional outcomes in adults who have had a stroke.

Objective: The objective of this study was to find evidence for common rehabilitation interventions to improve movement/motor and functional outcomes in adults who have had a stroke.

Methods: Medline Complete, PubMed, CINAHL Complete, Cochrane Database, Rehabilitation and Sports Medicine Source, Dissertation Abstracts International, and National Guideline Clearinghouse, from 1996 to April of 2016, were searched. From 348 articles, 173 met the following inclusion criteria: (1) published systematic reviews or meta-analyses, (2) outcomes target functional movement or motor skills of the upper and lower limbs, (3) non-pharmacological interventions that are commonly delivered to post-stroke population (acute and chronic), (4) human studies, and (5) English. Evidence tables were created to analyze the findings of systematic reviews and meta-analyses by category of interventions and outcomes.

Results: This rapid review found that the following interventions possess credible evidence to improve functional movement of persons with stroke: cardiorespiratory training, therapeutic exercise (ie, strengthening), task-oriented training (task-specific training), constraint-induced movement therapy (CIMT), mental practice, and mirror therapy. Neuromuscular electrical stimulation (NMES) (ie, functional electrical stimulation) shows promise as an intervention for stroke survivors.

Conclusions: Most commonly delivered therapeutic interventions to improve motor recovery after a stroke possess moderate quality evidence and are effective. Future research recommendations, such as optimal timing and dosage, would help rehabilitation professionals tailor interventions to achieve the best outcomes for stroke survivors.

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KEYWORDS

stroke; evidence-based health care; patient-centered care; review

Introduction

While the mortality rate from stroke has declined by 35% from 2001 to 2011, stroke remains the leading preventable cause of disability, leaving many stroke survivors with daily challenges [1], such as impairments in mobility, activities of daily living, house maintenance tasks, leisure activities, and stamina [2]. Stroke rehabilitation interventions are therefore critically important to maximize functional recovery and independence. Although an evidence summary of exercise therapy was published in 2005, it was not specific to stroke and was written in Dutch [3].

Among the many stakeholders interested in outcomes, clinicians and patients/caregivers seek trustworthy information about therapies (ie, evidence summary). Clinicians and policy-makers may not have the time to comprehensively research, given the rapid proliferation of research [4]. As patient-centeredness is increasingly adopted in clinical settings, it is important to not only respect patients' preferences, but to also facilitate patients' engagement and knowledge about their health condition [5]. Patients who have had a stroke may not be familiar with the terminology nor have the resources to efficiently search for evidence-based rehabilitation therapies to restore movement and functional outcomes. Recognizing that a thorough systematic review on this topic is beyond the scope of this article, we conducted a rapid review evidence summary to determine the level of evidence for common rehabilitation

interventions to improve movement/motor and functional outcomes in adults who have had a stroke.

Methods

Rapid review evidence summaries provide trustworthy information for broad questions to end users in a timely manner [6]. We specified the following inclusion criteria: (1) published systematic reviews and meta-analyses, (2) outcomes include functional movement or motor skills of the upper and lower limbs, (3) non-pharmacological interventions commonly delivered to poststroke population (acute and chronic), (4) human studies, and (5) English language. We excluded interventions that are not commonly delivered in postacute care settings, such as aquatic therapy and robotics. We sought Level 1 evidence from Medline Complete, PubMed, CINAHL Complete, Cochrane Database, Rehabilitation and Sports Medicine Source, Dissertation Abstracts International, and National Guideline Clearinghouse, from 1996 to April of 2016. Search terms included similar terms of the intervention, as well as *stroke*, *systematic review*, and *meta-analysis* (see Table 1). When questions arose about article inclusion or search terms, we discussed these items and rationales until an agreement was reached.

We screened 348 articles and identified 173 articles that met the inclusion criteria (see Table 2). Evidence tables were constructed to categorize and describe the results. After analysis, narrative summaries were written for each category.

Table 1. Search terms for interventions.

Category	Search terms used
Exercise–resistance training	Exercise, strength, resistance training, progressive resistance, physical activity, circuit training, cardiorespiratory, exercise therapy, function, intervention, mobility, motor, stroke, systematic review, evidence synthesis, meta-analysis
Constraint induced movement therapy	Constraint-induced, movement, systematic review, meta-analysis, stroke
Task-oriented training	Task-oriented training, task-specific training, stroke, systematic review, meta-analysis, repetitive task practice
Mirror therapy	Mirror therapy, systematic review, meta-analysis, stroke, motor
Neuromuscular electrical stimulation	Electrical stimulation, electrostimulation, electric stimulation, neuromuscular stimulation, systematic review, meta-analysis, stroke, motor
Mental practice	Mental practice, mental rehearsal, motor imagery, systematic review, meta-analysis, stroke

Table 2. Search results.

Category	Number of articles	Articles meeting criteria
Exercise–resistance training	165	55
Task-oriented training	54	35
Constraint induced movement therapy	34	26
Mental practice	22	14
Mirror therapy	19	12
Neuromuscular electrical stimulation	54	31
Total	348	173

Results

Interventions

In stroke rehabilitation, practitioners can choose among many rehabilitation interventions but this article will focus on interventions that facilitate functional movement and motor outcomes.

Based upon systematic reviews, common motor interventions include: cardiorespiratory training, therapeutic exercise, constraint-induced movement therapy (CIMT), task-oriented training or repetitive task practice, mental practice, mirror therapy, and neuromuscular electrical stimulation [7-9]. Interventions can vary by application, including method, therapist skill and familiarity of intervention, and amount of patient participation [10].

Cardiorespiratory Training

Cardiorespiratory training and aerobic exercise provide several health benefits to survivors of stroke. Two meta-analyses support aerobic exercise's positive effect on walking speed and walking endurance [11,12], but these effects do not extend to the Timed Up and Go (TUG) Test [11], Berg Balance score, Functional Independence Measure score [12]. Similarly, systematic reviews reported that gait-oriented cardiorespiratory training improves walking speed and tolerance [13,14], walking distance and peak oxygen uptake [15], and walking capacity [16]. Mixed training resulted in weaker effects on walking, and possibly balance [17].

While one systematic review reported an insufficient level of evidence for cardiovascular exercise's effects on disability, impairment, extended activities of daily living, and mortality, this review was published in 2003 and only included three trials [18]. Recent systematic reviews have concluded that cardiorespiratory training and exercise improve disability during or after usual stroke care [16,19] and improve health-related quality of life, respectively [20,21]. A review of 58 trials reported that cardiorespiratory training can produce moderate improvement on global indices of disability (standardized mean difference [SMD]=0.52, 95% confidence interval [CI] 0.19 to 0.84; $P=0.002$) [17].

Further research is needed to determine optimal dosing and long-term outcomes of cardiorespiratory training. From the 14 reviews found, cardiorespiratory exercise is effective for improving movement and health-related quality of life of individuals who have had a stroke [3,8,22].

Strengthening Interventions

Therapeutic exercise increases strength and activity [9,23], but the specific movement outcomes associated with exercise are unknown. Evidence syntheses from 23 reviews report the following benefits of exercise or strengthening interventions: (1) increased strength [23-25], (2) increased motor activity [23,24], (3) improved balance [26], (4) longer walking distance [15,27], and (5) faster walking speed [25,27-29]. Research suggests that circuit class training can improve walking distance [30], walking speed, and walking ability [31], even for individuals with chronic stroke [30]. However, passive

interventions appear less effective; one Cochrane review found little positive evidence for stretching, passive exercises and mobilization of the hemiplegic arm after stroke [32].

Based upon ten systematic reviews, progressive strengthening exercises are effective in improving leg strength and some aspects of gait performance [21,33]. Studies have shown that lower limb resistance training can improve comfortable gait speed and walking distance [34], as well as functional outcomes and quality of life [25]. However, there is insufficient evidence for lower limbs' effect on walking and balance [35] and pedaling exercise's effect on motor function [36]. A recent Cochrane review determined that using resistance training to increase walking speed is not supported by evidence [16]. Clinicians can reassure patients who have had a stroke that strengthening does not increase spasticity [23,24] or pain [24]. Among the four reviews examining the evidence for trunk exercises, two reviews concluded that trunk training exercises and lumbar stabilization exercises can improve trunk movement and dynamic sitting balance [37,38]. Aerobic exercise can improve balance of people with subacute and chronic stroke, whereas multisensory programs are less effective [26]. Moreover, balance training is feasible for people in a 1:1 model in the acute stage of stroke and either 1:1 or group therapy for those in the subacute or chronic stroke phase [39]. Exercise should be performed at least 20-60 minutes, 3-4 times a week, for 6-12 weeks. Evidence suggests that more is not necessarily better in the acute stage; exercising 90 minutes or more per day, 5 times per week may not be therapeutic [39].

Four reviews examined the effectiveness of bilateral and unilateral upper limb strengthening. A review from 2010 deemed the evidence for bilateral training as insufficient, when compared to placebo, no intervention, or usual care [40]. Van Delden and colleagues' meta-analysis reported that a marginally significant mean difference for perceived upper limb activity performance and quality of movement was found for those receiving unilateral training [41]. Although one review favored bilateral therapy's effect on upper limb function of adults with chronic stroke [42], the most recent review, examining functional task training, bilateral training with rhythmic auditory cues, and robot-assisted training, concluded that these therapy approaches produced results similar to usual therapy [43]. Thus, while bimanual training may improve proximal control, these benefits are offset by the reduced amount and quality of upper limb use from the participants' perspective [43]. Perhaps the conscious focus for unilateral training positively influences the participants' opinion of the affected limb's activity level and movement quality. Despite these findings, a Cochrane review highlighted the need for high-quality randomized controlled trials (RCT) to examine the effects of strength training [7]. In summary, bilateral training appears to produce results that are comparable to usual therapy, but unilateral training may produce better patient-reported outcomes.

Task-Oriented Training

Task-oriented training (ie, repetitive functional task training, task specific training) is a cost-effective intervention for individuals who have experienced a stroke [44]. Characterized by a composition of 15 components (eg, goal-directed,

functional, client centered, repetitious, context specific, progressive, and distributed practice), task-oriented training can be successfully applied when factors of intensity, duration of training, and the proper combination of specific components are incorporated [45]. Among these components, the use of “Distributed practice” and “Feedback” were associated with the largest postintervention effect sizes, and “Random practice” and “Use of clear functional goals” were associated with the largest follow-up effect sizes [45]. Interestingly, the number of components used during an intervention did not correlate with the posttreatment effect size.

Task-oriented training can improve gait, and can benefit people with chronic stroke [30,46]. Despite States and colleagues’ review concluding insufficient evidence for overground gait training [47], several systematic reviews support intensive repetitive task training’s effects on gait and gait-related activities [22,48,49]. While repetitive task training can produce significant gains in the movement performance of legs (eg, gait velocity, gait endurance, balance, Timed Up and Go Test), such effects do not extend to arm functioning [46,50]. One review suggested that task-oriented training may even be more effective than traditional therapies [51].

To improve aspects of walking ability, treadmill training can increase walking distance and maximum walking speed [52,53]. With regard to supporting body weight or not during treadmill training, it depends upon individuals’ walking ability. A recent Cochrane review found that individuals poststroke *who can walk* benefit more from body-weight supported treadmill training than people with stroke who aren’t able to walk, especially in walking endurance [54]. Veerbeek and colleagues’ review concurred with the use of body-weight supported treadmill training for improving walking distance and noted that electromechanical-assisted gait training with functional electrostimulation can improve maximum gait speed for dependent walkers in the early phase of stroke rehabilitation [21]. Generally, from the 21 reviews focused on walking, people with stroke can increase their walking speed and walking distance by treadmill training and body-weight supported treadmill training [21].

Task-oriented training’s effects on performance of daily activities appear to be minimal [49,52] or mixed [49,55,56]. For example, the effects of exercise on activities of daily living (ADL) were mixed; three meta-analyses reported a positive small to medium treatment effect on ADL [21,28,29], but another meta-analysis [57] and systematic review [18] did not find evidence for a favorable effect on ADL. High-intensity of practice results in improved quality of life and, as expected, leisure therapy improves leisure participation [21].

Outcomes from task-oriented training depend upon dosage and intensity (ie, dose x time). Evidence suggests that a higher dosage of task-oriented practice can improve arm functioning [10] and gait speed [21]. Jeon et al suggests training daily for at least two weeks for maximum progress [46]. Based upon 35 reviews, task-oriented training produces modest effects in outcomes related to leg functioning, but the evidence for positive effects on arm functioning is minimal [50].

Constraint-Induced Movement Therapy

We analyzed 26 systematic reviews and meta-analyses about CIMT, which involves restraining the functioning hand and encouraging the active use of the injured hand. Although the preponderance of evidence for CIMT is positive [9,22,58-63], the high intensity of functionally oriented task practice with the affected arm is difficult to implement because the protocol requires participation for over 90% of waking hours [64,65]. Moreover, CIMT is most beneficial for those who have at least 10 degrees of wrist extension, 10 degrees of thumb abduction/extension, and 10 degrees of finger extension in two other fingers [66]. Fortunately, modified-CIMT (mCIMT), with attenuated protocols or without a physical restraint, produces positive outcomes with less intensity [64].

Most evidence suggests that CIMT improves arm motor function [21,67], arm motor activity, [68,69] and movement quality [21] of the affected upper limb, even when the intensity is reduced and the duration is increased [70]. The schedule for modified CIMT (1 hour/day for 3 days/week for 10 weeks) is more feasible to implement [8]. For improved self-care, one review suggests a higher intensity of CIMT (at least 30 hours over 3 weeks) is beneficial [70], whereas another contends low-intensity mCIMT is effective [21]. In terms of quality of life, a majority of studies reported improved ADL [21], mobility, and participation [70,71]. Even more promising, CIMT appears to improve the amount and quality of active arm movements in people with chronic stroke [72].

Despite the positive findings of CIMT, some researchers have expressed some limitations of CIMT. Pulman and colleagues’ meta-analysis did not find significant improvements in ADL, hand function, and strength [73]. Additionally, a recent Cochrane review acknowledged that CIMT results in improved motor function and less motor impairment, but cautions that these small gains do not reduce disability [74]. While two reviews reported that CIMT’s effects (eg, arm motor activity) can be sustained for up to 6 months [9,68], Cochrane reviews have not found evidence for reducing disability several months after the intervention ended [74,75]. Acknowledging that CIMT can be useful in stroke rehabilitation, one review called for more research to determine if CIMT should be implemented as an adjunct therapy or as a replacement of traditional stroke therapy [76].

There are two issues to consider for clinical applications of CIMT. First, a meta-analysis comparing high and low-intensity CIMT in *acute or sub-acute stroke care* found that low-intensity CIMT may produce better movement and functional use of the affected upper limb [77]. While CIMT appears to produce better upper limb functioning than dose-matched interventions [69], only some of the CIMT RCT results produced minimal clinically important differences [78], raising questions about CIMT’s clinical efficacy [79]. More recently, Etoom and colleagues’ meta-analysis concludes that in comparison to other rehabilitation therapies, CIMT confers relatively small gains [80].

Mental Practice

In addition to physical rehabilitation interventions, mental practice can also improve movement and functional performance of patients who have had a stroke [9,81,82]. Mental practice demonstrates cortical activation patterns like those seen with actual movement, per functional imaging.⁸³ When combined with conventional therapies, mental practice improves recovery of both upper and lower limbs, as well as for reacquiring daily living skills [83]. Most systematic reviews focusing on mental practice to improve upper limb functioning, such as arm-hand activities, were positive [21,22,82,84,85]. Braun and colleagues reported that mental practice produced short-term gains in arm-hand ability as well as performance of activities [86]. However, Machado and colleagues' [87] meta-analysis did not find mental practice to be an effective adjunct therapy.

Adding mental practice to upper limb rehabilitation can independently increase functional recovery after stroke [85]. Cha and colleagues' meta-analysis calculated a medium effect size of .51 (95% CI: 0.27 to 0.75) for functional task training with mental practice during occupational and physical therapy in stroke rehabilitation [88]. Overall, evidence from 14 reviews suggests that mental practice is effective when paired with functional task rehabilitation for individuals who have had a stroke.

Mirror Therapy

Mirror therapy (MT) uses a strategically placed mirror to provide visual feedback of the unaffected hand's movements, creating an illusion that the affected hand is moving similarly. A majority of 12 systematic reviews reported positive effects of mirror therapy's efficacy for upper limb functioning [7,71,89,90], and two reviews found that MT's outcomes may be maintained for three to six months [9,91]. Only one review examined the effects of MT on lower limb functioning and found that MT is effective [92]. Whether MT can improve performance of activities of daily living is unclear; one review found MT to be effective based upon four studies [90], and another reported mixed results from three studies [85]. Questions about optimal dosage, timing, and application methods for people with varying stroke severity need to be answered in further research [91,93]. Capitalizing on neuroplasticity, mirror therapy appears to be an efficacious intervention for improving upper limb function after stroke, with moderate quality of evidence from a Cochrane review [94].

Neuromuscular Electrical Stimulation

From the 54 articles identified, we retained 31 reviews, and most (21) reported positive findings for neuromuscular electrical stimulation (NMES). In conjunction with functional activities, NMES addresses weakness, coordination, or spasticity, to improve function in poststroke population. NMES has a moderate treatment effect on activity when compared to training [95]. Proponents of NMES cite improved spasticity, range of motion [96], strength, and activity performance [97]. From Nascimento and colleagues' systematic review with meta-analysis, cyclical electrical stimulation increased strength by a standardized mean difference of 0.47 (95% CI 0.26 to 0.68) and this effect was sustained after the intervention with a small to medium effect size [97].

Evidence suggests that NMES can be effective when combined with other modalities for preventing and treating shoulder subluxation early poststroke [8,98,99]. Also, pairing functional electrical stimulation (FES) with an activity appears to be more beneficial than performing that activity alone (moderate effect size); a synergistic effect results when FES is used for improving upper extremity function (large effect size) [33].

NMES can also help improve lower limb motor performance and walking abilities, such as gait speed [100]. The orthotic effect of FES on walking speed was positive, with a pooled improvement of 0.13 m/s (0.07-0.2) or 38% (22.18-53.8) [101], and this effect has also been demonstrated for individuals with chronic stroke [102]. A recent systematic review examining the carryover effects of lower limb FES to motor performance concluded that FES produced therapeutic effects at the body function and activity levels, but there's insufficient evidence to ascertain the superiority of FES when compared to matched therapies [103].

Electromyogram (EMG)-triggered electrical stimulation has been shown to have mixed results. Chae asserted that repetitive movement training through transcutaneous cyclic and EMG-triggered NMES could improve stroke survivors' motor skills [104]. Meilink and colleagues' systematic review did not find statistically significant difference between EMG-NMES and usual care, citing a lack of rigor with the sampled studies, including non-randomization, small sample size, lack of blinding, and poor contrast to controls [105].

Evidence appraisers cite insufficient evidence for NMES' efficacy and low-quality trials [7,106-108]. A wide variety of therapy protocols, including duration of therapy [109], as well as heterogeneous samples, contribute to the difficulty in interpreting the evidence. For clinical applications, therapy practitioners should keep in mind that the evidence for NMES for individuals with chronic stroke is insufficient [99]. With more rigorous studies, NMES has the potential to improve functional motor abilities, especially in the acute phase of stroke recovery. Instead of offering FES to all poststroke, guidelines suggest considering electrical stimulation on a trial basis within the first two months poststroke to individuals who demonstrate muscle contraction but cannot move their limbs against resistance [8,110]. Overall, the evidence for FES to improve motor abilities is mixed, and therefore FES is a promising intervention.

Discussion

Principal Findings

No single intervention is superior to another in stroke rehabilitation to improve functional performance [7]. The following common movement-focused poststroke interventions have moderate evidence of effectiveness: cardiorespiratory training, therapeutic exercise, task-oriented training (task-specific training), CIMT, mental practice, and mirror therapy. While there are many systematic reviews and meta-analyses about movement interventions, the heterogeneity in samples (eg, acute vs. chronic), interventions (eg, timing, dosage, type), and outcome measures makes analysis of the

findings challenging. Combinations of evidence-based interventions across the postacute care continuum to address stroke patients' motor goals are considered standard care.

NMES is a promising intervention and more rigorous studies are needed to determine its effectiveness, particularly in the acute and subacute phases of stroke rehabilitation. Other interventions associated with stroke rehabilitation (eg, robotic therapy, aquatic therapy, and virtual reality and video gaming) are acquiring more evidence and some (eg, virtual reality) appear to be promising interventions.

To achieve the most recovery of activities of daily living, rehabilitation should be implemented early [7], and continuously across transitions and settings. Usual therapy approaches, including strengthening and functional activities, improved short- and long-distance walking, and thus strengthening interventions appear to be effective, even for individuals with chronic stroke [21,111]. Strengthening interventions may increase muscle functioning but may not necessarily translate to improved performance in ADLs, such as bathing, dressing, and meal preparation.

Recovery from stroke is a dynamic process and therefore rehabilitation professionals *craft* individualized therapy plans to maximize functional performance and participation. Highly effective therapy practitioners understand there is an *art and science* to providing therapy services and achieving excellent patient outcomes [112]. For example, a practitioner may recognize the science aspect by choosing low-intensity CIMT in acute or sub-acute stroke to improve functioning of the weaker arm [77], and use therapeutic rapport or humor to engage and motivate individuals with stroke to perform therapeutic tasks. Knowing when and how to deliver the most effective therapies, coupled with the collaborative and motivating aspect of therapy, will help therapists improve the movement outcomes and quality of care in stroke rehabilitation.

In patient-centered stroke rehabilitation, therapists and patients discuss goals, preferences, and concerns before co-creating treatment plans. Currently, therapists utilize a variety of interventions, considering the evaluation results, and patient/caregiver's concerns and aims. Consequently, rehabilitation professionals can best serve their clients with stroke by: (1) determining clients' goals and preferences, (2) thoroughly assessing their capacities and skills, and (3) selecting the interventions with the most relevant evidence that will enable clients to reach their goals.

In addition to the specific therapeutic interventions provided at different levels of care, other factors that influence stroke outcomes include: dosage and intensity of interventions, community support, education of staff and family, and caregiver competency. With regard to dosage, previous reviews have provided limited support for the assumption that a higher dose of exercise-based therapy improves motor recovery after stroke [28,113]. Generally, a dose of 30 to 60 minutes of therapy for five to seven days a week is optimal to improve performance [7,21]. Recently Veerbeek et al asserted that a higher intensity of practice (ie, 17 hours over 10 weeks) results in better outcomes at the body function level, as well as the activities

and participation level of the International Classification of Functioning (ICF) [21].

If only one intervention approach is being used for all therapy sessions or if the interventions cannot be named and explained by therapists, patients should seek additional information from their therapy practitioners. Sample questions patients can ask include: (1) Which intervention approaches are you using to achieve my goals? (2) What is the evidence for these approaches? (3) What can I do to maximize my recovery and motor outcomes?

Many patients poststroke may wonder if there is anything they can do at home to improve their motor outcomes. Coupar and colleagues' Cochrane review asserted that there's insufficient evidence to form any recommendations about home-based therapy programs to improve arm recovery [114], but recent reviews have cited positive effects of home-based therapies on functional performance [115,116]. Emerging technologies like virtual reality, robotics, and interactive video gaming hold great promise for increasing the dosage of movement-based therapies at home [116,117], which could potentially increase functional outcomes while containing costs.

Limitations

This evidence review did not include non-English systematic reviews and meta-analyses. We limited our search to the following databases: Medline Complete, PubMed, CINAHL Complete, Cochrane Database, Rehabilitation and Sports Medicine Source, Dissertation Abstracts International, and National Guideline Clearinghouse. Due to our focus on adults' functional movement of limbs and trunk, we excluded evidence that pertained to pediatrics and speech/swallowing. This review did not include robotic therapy, aquatic therapy, and virtual reality, because they are not commonly implemented in postacute care settings with most patients who have had a stroke. Some interventions lack the efficacy to improve activity performance (eg, neurodevelopmental treatment or NDT) but may be effective for other outcomes, such as improving muscle strength of the arm [9,21,118]. Lastly, we only included Level 1 published evidence and thus there is the possibility of recent RCTs not being included because of publication timelines.

Future Research

Practice guidelines recommend that stroke patients receive a minimal dose of active practice (ie, one hour each of physical therapy and occupational therapy) per day, at least 5 days per week [119]. Research is needed to identify not only the most effective combinations of movement-based interventions to deliver, but also the best critical window of time to deliver them. We need more research like Kwakkel and colleagues' meta-analysis, which reported that additional exercise therapy should exceed 16 hours within the first 6 months after stroke to achieve a statistically significant difference in ADL [29]. Such studies offer a clearer picture of the dose of an intervention, timing, and the anticipated functional outcome.

Moreover, therapy protocols need to be researched to increase our understanding of which subgroups benefit the most from certain interventions. In this era of personalized medicine, there may be subsets of stroke survivors who would benefit from a

certain therapeutic cocktail of interventions across settings to achieve maximum functional recovery. Multi-site studies and registries could help add to existing databases by collecting data about demographic variables, stroke types, costs, and functional movement outcomes.

Another issue is the wide variation of outcome measures used to measure functional movement. Informed discussions between researchers and clinicians could not only stimulate and focus rehabilitation research, but also pave the path towards attaining consensus about best outcome measures and intervention methods for stroke survivors. Consensus about outcome measures and which interventions to study during the phases of stroke recovery could facilitate comparative effectiveness research.

Additionally, high-quality RCTs are needed to determine if poststroke interventions targeting body functions lead to

improved activity and participation [120]. Finally, we need rigorous longitudinal studies to examine cost-effectiveness and the effects of strength training on activity and participation, and to determine to what extent any gains are sustained.

Conclusions

Patients and rehabilitation professionals may be more reassured that the following interventions possess moderate evidence of effectiveness: cardiorespiratory training, therapeutic exercise, task-oriented training, constraint-induced movement therapy, mental practice, and mirror therapy. More research is needed to determine the optimal timing and dosages of motor interventions, as well as the effectiveness of neuromuscular electrical stimulation. Movement outcomes are influenced by many variables, such as stroke characteristics, intensity, social support, as well as patients' preferences and goals.

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Conflicts of Interest

None declared.

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Abbreviations

- ADL:** activities of daily living
- CIMT:** constraint-induced movement therapy
- EMG:** electromyogram
- FES:** functional electrical stimulation
- ICF:** International Classification of Functioning
- mCIMT:** modified-CIMT
- MT:** mirror therapy
- NMES:** neuromuscular electrical stimulation
- RCT:** randomized controlled trial
- SMD:** standardized mean difference
- TUG:** Timed Up and Go

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Viewpoint

Patient and Family Involvement: A Discussion of Co-Led Redesign of Healthcare Services

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Abstract

The involvement of patients and their families in the redesign of healthcare services is an important option in providing a service that addresses the patients' needs and improves health outcomes. However, it is a resource-intensive approach, and it is currently not clear when it should be used, and what should be the reasoning behind this decision. Some health systems of international standing have created a patient engagement program as a selling point. This paper discusses how co-led redesign can be beneficial in improving health service and more effectively engaging patients. Potential barriers for patient involvement are discussed. Patient involvement can be integrated into the health system at three main levels of engagement: direct care, organizational design and governance, and policy-making. The aim of this paper is to describe how co-led redesign is compatible with different levels of patient involvement and to address the challenges in delivering a co-led redesign in healthcare. Co-led redesign not only involves the collection of quantitative data for assessing the current systems but also the collection of qualitative data through patient, family, and staff interviews to determine the barriers to patient satisfaction. Co-led redesign is a resource-rich process that requires expertise in data collection and a clinical group that is devoted to implementing recommended changes. Currently, a number of countries have utilized co-led redesign for many different types of healthcare services. Resource availability and cost, process time, and lack of outcome measures are three major limiting factors.

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KEYWORDS

co-led redesign; health care; patient and family engagement; patient involvement; rural health

Introduction

The involvement of patients and their families in the planning and development of health care services has been shown to improve the health and quality of life of patients [1]. Patient and family engagement has been defined as “a relationship between health care providers working together to promote and support active patient and public involvement in health care and to strengthen their influence on health care decisions at an individual and collective level” [2]. Patient and family involvement includes the feedback and experiences from patients and their family members and caregivers. They have experience,

expertise, insights and valuable perspectives that are useful in bringing about changes in health care regardless of whether their own experience was positive or negative. Patient engagement, including partnerships, transparency, and information sharing between providers and patients, can be applied to decision making at an organizational level. System modification is required to ensure that patients and families have a voice in planning organisational strategy and in designing changes and improvements in patient care [3].

A number of studies have demonstrated the importance of involving patients and their families in health care redesign initiatives through patient satisfaction surveys or interviews and

mapping of patient journeys. Patients in a study of stroke services in the United Kingdom reported high levels of satisfaction with inpatient care [4]. A substantial proportion of patients and their families also reported dissatisfaction with the lack of involvement in decisions [5]. A different patient journey study in the UK [6] showed that four main themes emerged from questioning patients and their families about their in-hospital experiences; (1) information provision, (2) staff attitudes, (3) availability of care, and (4) considering the whole person in context. This paper defines patient involvement in health care and demonstrates the current state of patient and family engagement in the redesign of clinical management for health care services. This paper also highlights the barriers to achieving adequate patient and family engagement in a health care service redesign process.

Patient Involvement

In 1988, the term patient-centered care was used to call attention to the need for clinical staff and health care systems to shift their focus away from diseases and back to the patient and family [7]. This movement was designed to stress the importance of understanding the patient experience and delivering more effectively on patient needs, including decisions about their care and treatment, diagnostic tests, screening, and medications. This term still refers to a focus on patients, however, it does not necessarily reflect that patients should also be involved in their care or at what level. Patient and family engagement or involvement is now a main driver for improving quality in health care. This engagement can range from consultation to partnership and from a limited decision-making role up to a shared decision-making role. Patient and family engagement has many forms and can occur for a number of reasons. Three levels of engagement for patient involvement, identified by Carman and colleagues [8], are direct care, organizational design and governance, and policy making. This framework is the basis for defining patient and family involvement and co-led redesign in this paper.

Direct Care

Dieppe and colleagues [9] state that the clinical encounter, the point at which health care professional and patient interact, is “at the heart of health care.” Patient involvement, from a direct care viewpoint, involves including the patient and their family in the decisions that are made about their diagnosis and treatment. It is defined as integrating the patients’ values, experiences, and perspectives in relation to prevention, diagnosis, and treatment [9]. The involvement of patients and their family members in improving the quality of health care has been considered to be a democratic or ethical requirement as patients indirectly pay for services through taxation (in some countries, including Australia) and therefore should have a right to influence how they are managed [10,11]. In the United States, however, employers are often responsible for paying health insurance premiums for their employees’ hospital treatment, often with a copayment from the employee, as per the market-based health insurance system. In this instance, patient involvement might seem more of a priority as there is direct

payment for the services utilized. Other ethical considerations, as described by Elwyn and colleagues [12], include individual self-determination and the idea that clinicians need to support this. Self-determination, in the context of shared decision-making and patient and family involvement in health care, pertains to an intrinsic human tendency to preserve one’s own well-being [13], which is something that not all patients or families exhibit. At the direct level, patient and family involvement could mean simply providing patients and their family members with information or involving them actively in setting goals or making decisions about their care.

Information provision was a major theme identified by Morris and colleagues [6] during a patient journey study. Providing patients, and their families and caregivers, with accurate and suitable information is an important component of direct care. Actively involving patients and their families by ensuring they receive and understand information about their condition including treatment has been shown to improve quality of life significantly when compared with patients who did not actively receive this information [14-17]. The Royal Children’s Hospital (Melbourne, Australia) developed a policy that defines patient and family-centered care, including sharing of information, involving the patient and family in decision making, and sharing the provision of care. This direct style of patient involvement is certainly family- and patient-centered but the development of clinical procedures, pathways, and mode of service delivery are still decided by clinicians, with little patient or family input. Currently, many hospitals have policies and procedures that encourage patient and family involvement at the direct level of engagement, but not necessarily in the redesign of the health care services they utilize.

It has been suggested [18] that the most important attribute of patient-centred care is the active engagement of patients when health care decisions must be made. Graffigna and colleagues [19] developed the Patient Health Engagement model which provides an overview of patient engagement. It consists of four stages. Each level addresses a significant stage in the patient journey where the patient becomes a “co-constructor of their health and capable of self-management.” Engaging patients in this way, to allow a sense of control and understanding of their condition, has been shown to have a positive effect on patient satisfaction. It also reduces depression [20]. However, a study by Sommers and colleagues [21] found that some patients would prefer that their health care providers just tell them what to do rather than engage in shared decision making. This suggests that some patients may be less likely to benefit from more collaborative levels of participation. This external aspect of control [22] is a conundrum for proponents of patient involvement, as lower socioeconomic status (SES) patients tend to have less internal control over health. Therefore, while higher SES patients are more likely to take advantage of patient involvement systems, the need is for the lower SES patients to be more involved, and take more control of their health.

Organizational Design and Governance

Patient involvement in organisational design and governance provides an opportunity for patients to partner with health care

providers in planning, delivering and evaluating health care. This encompasses involvement in the design of the health care facility, through to assisting with hiring and training staff. A review of patient and public involvement in health care in the UK was conducted by Mockford and colleagues. They divided the impact of this involvement into service planning and development, information development and dissemination, and changing attitudes of service providers and users [23].

Other forms of patient and family engagement for delivery of improved organizational goals include participation on consumer committees, patient satisfaction surveys, participating in focus groups, and patient and caregiver representation on planning and development boards/panels. Patient involvement at an organizational level also includes participation in quality improvement opportunities. Feedback, including complaints and compliments, can be utilized by management teams to improve the future design of health care services and make changes in governance and policy. Reid Ponte and Peterson [3] suggest that the principles of partnership, transparency, and information sharing must guide the interactions between providers and patients and their families at the bedside, and then be applied to the organizational level.

Policy Making

Patients, or members of the public, can collaborate with representatives from health care facilities to make decisions about how to shape health care policies and set priorities for the use of resources. Described in the UK as a “remarkable experiment in democratic practice” [24] is a form of patient engagement known as citizen juries. A citizen jury consists of a defined number of carefully selected ordinary citizens who address questions about policy and planning in health care in a primarily advisory role. They are provided information from “witnesses” in their quest to reach consensus around specific health care issues. Although the benefits of a citizen jury include information, time, and independence [25], this process does not provide a real-life account of the experience of the current health care system.

Co-Led Redesign

Co-led redesign can be defined as “the development and implementation of health care services based on both a clinical and patient perspective and experience or experience-based design.” It involves clinical engagement, patient and family engagement, shared decision making and a thorough analysis of the current systems and expected benefits of new, improved systems. Co-led redesign occurs at all three levels of engagement in differing capacities. In the patient journey clinical redesign process [26], a co-led redesign process, the direct engagement occurs via a systems analysis, performed from the perspective and experience of the patient, and their families. It also includes the front line health care staff who are critical to improving the clinical practice. Organizational design and governance is assessed through a quantitative analysis of data, linked to direct engagement information to form new policy and practice. This evidence-based approach utilizes quantitative and qualitative data to inform the decision makers, at all levels, how to allocate

resources and structure health care service provision. Patient involvement in health services redesign is based on the premise that involving patients leads to more accessible and acceptable services and improves the health and quality of life of patients [1]. Mixed method research (combining quantitative and qualitative analysis) can capitalise on the strengths of each approach. This includes corroborating findings, generating more complete data, and using the results from one method to enhance the insights from the other. In the UK, government policy states that “involving patients and the public isn’t always easy and can take time but, done well, has been shown to be highly effective in developing services that better meet patient needs and lead to better health outcomes” [27]. There is evidence to suggest that patient engagement in the redesign of health care services is linked to fewer adverse events, better patient self-management, fewer diagnostic tests, decreased use of health care services, and shorter lengths of stay in hospitals [28]. Experience-based design is a user-focused design process with the goal of making user experience accessible to allow design of a better patient and staff experience [29]. Co-led redesign places the experience goals of patients and their families and staff at the centre of the design process. It creates a partnership with the patients, families and staff, and promotes shared leadership and decision making.

Use of Patient Engagement within Redesign and Co-Led Redesign Models

Instances of direct engagement, organisational design and governance changes and policy making can be shown through varying types of patient involvement initiatives locally and globally. A national survey of hospitals in the United States [28] reports that of the 1457 hospitals that responded, 7% include patients and family members in the education and content development when training clinical staff, 21% had a patient and family advisory council that had met within the previous 12 months and 23% had patient and family advisory councils [28]. This type of patient and/or family involvement is not necessarily for health care redesign purposes, although it does provide an opportunity for patients and families to give input regarding various hospital activities. This suggests that while patient and family engagement is occurring, the level of participation is inadequate for a patient journey co-led redesign process, which requires more in depth, personal patient and family involvement. Although this information is valuable and the economic impact of utilizing patients and their families in the redesign of health care services has been shown to be a limiting factor, particularly when interviewing individual patients and families is chosen research method [23].

Co-led redesign incorporates the patient and family feedback and suggestions on how to improve current services based on previous experience in a particular health service in conjunction with other research methods. The qualitative method of interviewing patients and their families allows them to identify gaps and strengths of the system and influence the redesign of health care services. Patient and family feedback is utilized to directly influence changes to be made to existing services or to the development of new services. A systematic review of

involving patients in planning and development of health care services [30] reported that patients who participated in these initiatives welcomed the opportunity and that their self-esteem improved as a result of their contribution. This review suggests the most frequently reported effects of involving patients in developing or improving health services include making services more accessible through simplifying appointment procedures, extending opening hours, improving transport to treatment units, and improving access for people with disabilities. Patient involvement may take place via face-to-face meetings, patient representation in planning meetings, group interviews, written surveys and consumer boards [30].

Collection of qualitative data in co-led redesign, through direct engagement methods, patient interviews, or other interactive forums, is time consuming and resource rich and its subsequent analysis is much the same. Qualitative methods generate a substantial amount of data, it is suggested that just 20 one-hour interviews can generate up to 400 single spaced pages of transcripts [31]. Thematic analysis has been shown to be effective in identifying gaps in service, areas for overall improvement and barriers to effective service delivery despite the timely analysis process [32]. Identifying themes and patterns from the experiences of patients and their families ensures a comprehensive view of the overall service but involves a number of time consuming components. Themes are defined as units derived from patterns such as conversation topics, vocabulary, recurring activities, meanings, feelings, or folk sayings and proverbs [33] and are often determined from recorded interview transcripts. These can then be divided further into sub-themes for identifying further patterns in data. Following theme definition, a literature review should be performed to validate the argument for the choice of themes (and sub-themes) in order to build a report, or story that highlights the patient perspective on the health service. Broad outcome measures for quantitative analysis, such as length of stay and readmission rates, are useful across many conditions. However, developing condition-specific measures, (effective interventions to improve the quality of care for qualitative analysis in co-led redesign becomes a time consuming and difficult process. Both methods of research have benefits in co-led redesign. Quantitative research counts occurrences (eg, prevalence, frequency), whereas qualitative research, following a thorough analysis, can describe the complexity, range of or breadth of occurrences and generate hypotheses about a particular phenomenon [31].

New South Wales Health (Australia) and Flinders Medical Centre (South Australia) underwent a major clinical services redesign program between 2002 and 2005 [34] utilizing the patient journey method. Mapping of the current patient journey was performed by involving all staff members as well as interviewing patients and their families about their experiences within particular health services. This provided an avenue for patients and their families to reflect on the service, provide feedback about potential improvements, and gave them an opportunity to speak openly about strengths and weaknesses of the overall health care service. The clinical services redesign in New South Wales is ongoing and is delivered as part of the NSW Agency for Clinical Innovation Program [35]. The benefits of the Flinders Medical Centre redesign include the stabilizing

of staffing, reductions in the numbers of adverse events throughout the hospital and reduced length of stay for medical patients admitted as emergency cases [34]. In the United Kingdom, the involvement of patients and the public in shaping health care is well established in National Health Service (NHS) policy and reinforced by a government that is committed to empowering individuals to play a greater role in their own health care [36]. The type of redesign developed within the NHS has been utilized for a number of health care services including prostate cancer, acute coronary syndrome, cholecystectomy and head and neck cancer. The prostate cancer redesign resulted in changes that could not have occurred without using a co-design process with [patient] interviewees, such as through the support group that was developed. Data from the prostate cancer redesign indicated that appointments were not coordinated for the patients and their family. A solution that was proposed was a one-stop-shop for all diagnostic tests, but the men and their wives [from the support group/ from the interviews] considered that all of the tests being done on the one day would be too much. [26].

Co-led redesign has also been successfully implemented in services in Sweden, where patients with diabetes were consulted formally about the existing diabetes management in primary care plans. This led to changes to the organization of care and in the type of information provided to patients utilizing this service [37]. Between 2004 and 2008, a patient-centered method to redesign patient care delivery was developed and refined at the University of Pittsburgh Medical Center (US) as a means of improving patient care experiences and exceeding the needs and desires of patients and their families [38]. This was a co-led redesign process involving the selection of a particular patient experience (health service), establishment of a patient and family-centered care experience group, the mapping of the complete patient journey (surveys, storytelling, patient shadowing, and family experience), and the involvement of all staff in the care experience. This initiative resulted in a dramatically improved service and outcomes without increasing cost. It also eradicated silos that are often seen in hospital systems. Results include a 14% significant increase in patient satisfaction in the emergency department and a 13% significant increase in patient satisfaction in the general trauma ward [38]. The savings in this study can be attributed to the development of patient and family experience initiatives based on the timeline of implementation and evaluation. This study also resulted in a decrease in staff turnover of 66% over three years, as well as an annual saving of \$5,000 in one inpatient unit by changing the late food-tray menu and process.

Barriers to the Use of Co-Led Redesign

General Barriers

While co-led redesign has many benefits, it also has some limitations. The staff resources required to complete the quantitative and qualitative analysis adequately on behalf of the health service can create delays and can be quite costly, resource-wise and financially. Completion of this work by health care staff may also create staff stress and anxiety if they feel inexperienced and lack time and resources. Co-led redesign

involves the establishment of a patient journey group to oversee the process and take on the responsibility of ensuring that the redesign project is understood and working well. The patient journey group comprises a number of roles including a chair, lead, and a clinical champion, all coming from within the organization and a patient journey facilitator who is not a part of the clinical team [26]. Knowledge management is a key part of the co-led redesign process. Regular meetings and communication of research findings, progress reviews, maintaining focus, and delivering on the expectations of the redesign are important in ensuring overall success. Again, time and resources often don't allow key players to be fully invested in co-led redesign, despite their intentions for change. While a number of patient and family engagement initiatives have been developed in many health care organizations outside of co-led redesign, they often lack clear guidelines and fail to reveal an evidence-base to explain or support the approach [39].

Some potential barriers for overall consumer and patient involvement in health care and general redesign initiatives were identified by Nilsen and colleagues [40]. Health professionals often view themselves as authorities. People may believe that involving patients in policy, research, and practice increases costs and causes delays. They may also fear that patients may have biased views that interfere with the "academic impartiality" of knowledge development [41]. Another factor found to impede meaningful patient involvement is organizational and professional resistance to change or learning something from health service providers [42]. Evidence suggests that a blame culture within health care organizations prevents staff from being open and sharing their views [43-46]. Managerial interest is often focused on budgets and targets and achieving status rather than on patients and their families. There is also evidence to suggest that staff shortages, lack of time and resources, poor communication, and fragmented ways of working continue to affect both patient and staff experiences adversely [43,46-48]. Issues based on the patient and staff experience with care, and the delivery of care in general, are important for understanding potential issues within co-led redesign. It is imperative to ensure that the patient and family experience and story is not demeaned by a lack of managerial support for co-led redesign, or that a focus on administrative targets prevents the full involvement of patients and their families.

Health Literacy

Another major limiting factor in direct engagement of patients and their families is low health literacy, particularly in areas known to have a low SES. Initial and ongoing patient participation in the qualitative component of co-led redesign is dependent on a number of patient-specific issues, as identified by Jordan and colleagues [49]: the ability to identify and understand health messages, having access to information and services, and possessing the skills to decide which information is useful. A key limiting factor for active patient participation in developing and building relationships with health care providers is health literacy of the patient and their family. Education and health literacy potentially limit a person's ability to be involved in decisions about their health [50] and the health care of their families. Greater involvement places an increased demand on each patient's literacy skills in order to understand

complex health information and articulate their preferences and their experiences [50]. Co-led redesign relies on information directly from the patient and their families through directly asking the patient about their experience. Health literacy levels have been shown to influence this information. In a study by Smith and colleagues [50] patients with a lower health literacy level reported that they were not interested in trying to understand the "mechanics or you know, pros and cons". These patients were more interested in having their doctor take the lead and offer a definitive decision. Patients with a higher health literacy, reported seeking independent knowledge around their condition, although still respected their doctor's expertise. While not directly related to health literacy it is also important to consider and establish a method of meaningful communication with patients and family members who may have communication impairments such as aphasia, deafness or some forms of mental illness. To overcome this issue, it is important to recognize the differing health literacy levels in individuals and present the information accordingly. In order to empower patients and their families who have low health literacy levels, and give them the opportunity to participate in co-led redesign, all contact should be made personally and in a manner that creates an environment where questions are welcome and information can be understood.

Poor health literacy and low SES tend to go hand in hand, as does chronic disease and low SES [51][52]. Gaining informed consent from this group in co-design is a challenge, but they are the very people who need to take part. Informal approaches to such potential participants, such as phone calls, can open up fruitful participation, as opposed to a formal letter. Focus groups may be a good way to gain views from higher SES patients with good health literacy, but can be very threatening for other groups. The venue for an interview may provide a solution by changing the power relationship. For example, offering to do the interview in the patient's home, with supportive family members or friends present, may convince him to consent. The style of the interview is important in terms of using the appropriate level of English and being clear about the concepts concerned. An interviewer who creates an environment in which the participant knows that there is no right or wrong answer, and that his or her views are valued, is crucial.

Complaints Versus Feedback

Another barrier to successful patient led initiatives around improving health care services, identified by Mead and Bower [53] is the tension between the aims and priorities of health practitioners and those of the patients and families. This conflict has been identified as a limitation in the organisational design and governance engagement level as well as from a policy making perspective. If there is a specific issue or complaint from a patient or family member, this may be the only focus of their involvement and they may become distracted from the overall feedback process. The involvement of patients and families with their own agendas for taking part in the research project may be counterproductive as their attention is on one aspect of service and they may fail to become engaged in the overall feedback journey. An interviewer can ensure that the direction of the conversation remains focused on experience feedback and relevant information by using a semi-structured

questioning technique, allowing the interviewer to bring the conversation back when required.

Conclusion: Future of Co-Led Redesign

Co-led redesign has been shown to have a number of benefits over traditional health care redesign initiatives, most notably, adding the patients' (and their family's) perspective to particular health care services. This information not only provides another source of evidence to build a case for redesign, but it also ensures that patients and their families are able to share their unique experiences, are represented, and feel involved in the improvement of the delivery of their health care services. There

are many successful redesigned services around the world that are a result of a co-led approach. This suggests that this method may be a major option for the future of clinical redesign.

But, without serious investment in an infrastructure for co-led redesign, as well as a commitment from its leadership and management, it is not possible for most health systems to adopt co-led redesign as their standard approach. Indeed, even a local single service co-led redesign is resource intensive, requiring a clinical team that is open to being informed in this way. It can be a challenge to find staff or university partners with the skills to bring in the patient voice. Clearly, there is much more work that needs to be done to fully develop a co-led redesign model for health care.

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Authors' Contributions

SJP contributed 80% to the manuscript including the full first draft and changes based on feedback. SC contributed 20% to the writing of the manuscript through providing direction and new ideas, editing and feedback.

Conflicts of Interest

None declared.

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Original Paper

Participant-Partners in Genetic Research: An Exome Study with Families of Children with Unexplained Medical Conditions

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Abstract

Background: Unlike aggregate research on groups of participants with a particular disorder, genomic research on discrete families' rare conditions could result in data of use to families, their healthcare, as well as generating knowledge on the human genome.

Objective: In a study of families seeking to rule in/out genetic causes for their children's medical conditions via exome sequencing, we solicited their views on the importance of genomic information. Our aim was to learn the interests of parents in seeking genomic research data and to gauge their responsiveness and engagement with the research team.

Methods: At enrollment, we offered participants options in the consent form for receiving potentially clinically relevant research results. We also offered an option of being a "partner" versus a "traditional" participant; partners could be re-contacted for research and study activities. We invited adult partners to complete a pre-exome survey, attend annual family forums, and participate in other inter-family interaction opportunities.

Results: Of the 385 adults enrolled, 79% opted for "partnership" with the research team. Nearly all (99.2%) participants opted to receive research results pertaining to their children's primary conditions. A majority indicated the desire to receive additional clinically relevant outside the scope of their children's conditions (92.7%) and an interest in non-clinically relevant genetic information (82.7%).

Conclusions: Most participants chose partnership, including its rights and potential burdens; however, active engagement in study activities remained the exception. Not surprisingly, the overwhelming majority of participants—both partners and traditional—expected to receive all genetic information resulting from the research study.

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KEYWORDS

partnership; exome sequencing; genome sequencing; return of results; participant engagement

Introduction

Researcher-Participant Partnerships: A Tough Walk to Walk?

Instantiation of reciprocal partnerships between researchers and participants has long been difficult to achieve, given: (1) historical norms of asymmetric researcher and participant relationships; (2) regulatory and policy disincentives to open communication (eg, Health Insurance Portability and Accountability Act [HIPAA] privacy provisions as applied to research participants; liability fears); and especially (3) practical and resource challenges to researchers [1]. Consequently, with few exceptions, one could argue that most unfettered big-data researcher-participant engagement has tended to happen in settings outside of academic medical centers (eg, Open Humans, PatientsLikeMe, Genetic Alliance) [2-5].

Partnership with patient participants in genomics-based research is embedded in the All of Us Research Program (formerly the Precision Medicine Initiative) and cited as an important component of open research communication that enables autonomy and choice in participation in long-term studies [6,7]. It was discussed as a salutary outcome in the 2015 Notice of Proposed Rulemaking that led to the reform of the Common Rule, the multi-agency framework that governs human participation in US research [8,9]. On a grassroots level, research participants have expressed both a strong willingness to share data derived from their samples and/or personal information and a desire to receive individual results from researchers [10].

Rule changes to HIPAA and the Clinical Laboratory Improvement Amendments of 1988 (CLIA) that took effect in 2014 suggest that research participants now have broad data access rights from any laboratory that behaves as a HIPAA-covered entity [11]. While some have regarded this development as “troubling,” we view it as an opportunity to begin to realize the aspirational notions of partnership expressed by All of Us, broadly by patient-centered research (eg, the Patient-Centered Outcomes Research Institute) and by participants themselves [12].

Research-based Exome Sequencing and Functional Analysis in Undiagnosed Children: A Partnership Test Bed

The precipitous decline in the cost of whole-exome sequencing (WES) has made its use as a diagnostic approach increasingly commonplace [13-14]. Over the last several years, the power of WES to end diagnostic odysseys and, in some cases, to alter the course of clinical care, has been supported with an increasing number of examples [15-16].

Since 2011, our research group, the Task Force for Neonatal Genomics (TFNG), has received referrals from more than a dozen clinics within the Duke University Health System and

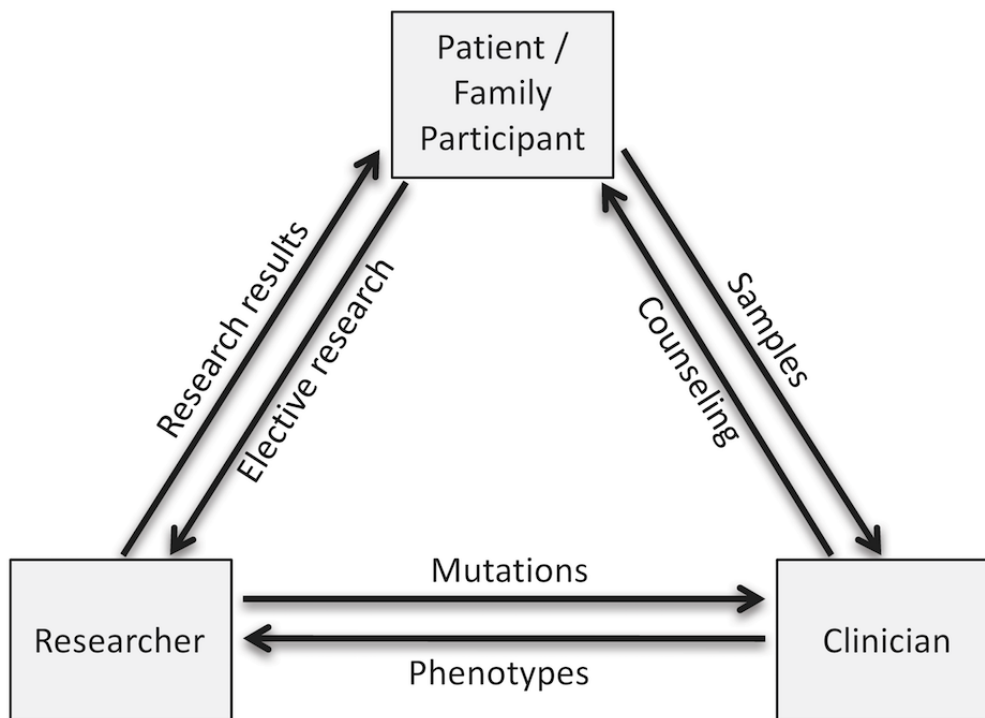
elsewhere across the US for young children with congenital structural anomalies likely attributable to a genetic cause. Pediatric diagnostic challenges arise among many specialty clinics, with only a small proportion referred to medical genetics [17]. By engaging directly with the specialty clinics, in many cases prior to a genetics referral, we could begin exploring possible genetic causes and engage with families who might otherwise not have been considered for exome sequencing.

Here we describe the efforts to increase participant involvement in the research and subsequent effects; the enrollment processes will be described later in the paper. In general, if clinic referrals met our consensus inclusion criteria, we enrolled trios (or quads, etc, if more than one child were affected) of biological parents and children with undiagnosed conditions for research-based WES.

We exome-sequenced each individual (both biological parents and affected children), and filtered candidate alleles using published and in-house algorithms. Candidate causative alleles were confirmed by Sanger sequencing in all enrolled family members, then parsed further through literature searches and, when possible, modeled in zebrafish to test causality of suspect alleles [18-23]. Use of animal models in combination with WES is a flagstone of this research project and distinguishes our study from standard clinical exome sequencing. In some cases, families pursue clinical exome sequencing as well; in other cases, families have already received inconclusive clinical exome results and have enrolled in our research study for data reanalysis and the development of zebrafish assays. As of July 2016, we had enrolled 225 families, and returned results on 52 probands. The remaining families are currently in the analysis pipeline in our study.

We define partnership as a reciprocal exchange of information and communication to the benefit of all parties and participation as partners as an educative dividend of that exchange [24]. In our relevant research context – that is the investigation of a genetic cause to a child’s medical condition – the information exchange can be mutually beneficial to families of the child, researchers, and the health care providers caring for the child. In family-based research on a rare condition, the family-specific research data may be of personal utility (if not clinical utility) beyond use in aggregate research studies. From the outset of this project, our mission has been to engage with families seeking a genetic etiology for their child’s medical condition near the onset of their diagnostic odyssey, or at least from the point at which they are referred to Duke University Medical Center. For this reason, we designed our protocol to enable return of genetic information relevant to the child’s condition, whether it was conclusive or not. To guide the choices of families potentially wary of engagement with the clinicians and research team and to further this objective, we developed a “partnership” model for families.

Figure 1. The partnership model of interchange of information fosters long-term prospective communication of phenotype, genetic risk, and interpretation of research results.



To that end, we developed three ways to engage family participants. First we developed a consent process that would allow partnership with families that would enable open communication between the research team and the participants, rather than relying upon the clinical referral team to transmit and receive information from individual participants (Figure 1). This partnership model is an option presented in the initial consent form. Second, we developed an online survey to gauge partner-participants' pre-exome interests in aspects of the research, including return of research data. Third, we hosted three annual family forums with the aim of bringing participant families together with the researchers and clinicians managing their research exomes. Here we present our findings on partner participation among our exome research enrollees and their pre-exome interests in receiving genetic research data.

Methods

Participant Enrollment Process

All study materials and protocols were approved by the Duke University Health System Institutional Review Board (DUHS Pro00031066). Pediatric and prenatal probands were referred from clinics across Duke, primarily pediatric urology, neurology, cardiology, craniofacial, bone marrow transplant, maternal-fetal medicine, and neonatology. Consent materials were available in Spanish. Genetic counselors reviewed case medical histories and discussed the phenotypes of individual cases with referring clinicians. The study team prioritized enrollment of cases that: (1) were suspected to be genetic (eg, no known maternal confounding or environmental factors; actual or possible recurrence within the family); (2) had no prior molecular genetic diagnoses as determined by clinical genetic testing (eg, WES,

panel sequencing, karyotyping, or array analysis); and (3) had phenotypes for which an anatomical surrogate could be modeled in zebrafish. Cases that did not meet pre-determined criteria were excluded by consensus or voted upon by study team members. Once the team decided to include a case, a health care professional known to the family introduced them to a research genetic counselor who then presented the study and consented willing family members, which, at minimum, included a trio of both parents and an affected child. The team required the availability of both biological parents to qualify for enrollment unless there were either multiple affected individuals or multiple generations of available family members.

Consent Process

Genetic counselors described the scope of the study and the potential for obtaining exome-based research results, including variants directly relevant to the affected child's condition, variants clinically relevant to the child or parent, and variants unrelated to the condition and/or not clinically relevant. The consent process included options for receiving directly relevant results, as well as clinically relevant results (ie, additional or "secondary" findings). The TFNG research protocol permitted return of clinically relevant results only; however, the consent form asked participants for their potential future interest in receiving results that were not clinically relevant, and a subsequent amendment to the protocol allowed for return of raw sequence data. Finally, the consent form provided an option to be a "partner" in the study or a "traditional participant" (relevant extracts of consent form available as supplementary information). Partners and researchers could communicate in an unfettered way via email; partner participants agreed to be

re-contacted for surveys and invitations to Task Force events such as seminars and participant gatherings.

Survey Development and Collection

Survey questions and design were developed in consultation with internal and external content experts, including genetic counselors, clinical geneticists, and genetic/genomic researchers. The survey questions (available as supplementary information) contained four main sections: (1) demographics (age; relationship to child; age of affected child); (2) experience in clinical or research-based approaches to identify an underlying cause of the child's/children's condition (length of diagnostic odyssey; number of specialists sought; length of time in research); (3) perspectives on research participation (rating scales); and (4) expectations for research (yes/no questions). In total, the survey consisted of 16 questions formatted as multiple-choice or nine-point sliding scales (ie, rate from 0-10). The survey was designed and distributed using Qualtrics software (Qualtrics, LLC, Provo, UT). Only adult parent participants who consented to be partners were eligible for the survey. Partner-participants who did not provide email addresses were excluded. The survey was conducted in English; participation was voluntary and anonymous.

Eligible participants were emailed an introduction to the survey, including a consent document and a link to the anonymous survey, within a month of the family's samples having been collected and the WES pipeline having been initiated. A single reminder email was sent about a month after the initial introduction. Because responses were anonymous and not linked to family identifiers, in some cases both a father and mother may have responded to the survey. In some cases, individuals were re-consented on the exome protocol at a later date to expand their participation options, so those samples would have been undergoing analysis for longer than a month.

After survey data collection, we conducted quality-control checks to assure that value ranges and missing data codes were valid. Data were analyzed for possible sources of response bias including inspecting individual responses for extreme bias and evaluation of consistent data trends over time. Responses were summarized using frequency distributions. For sliding scales, the mean was used with standard deviations (SD) to demonstrate response clusters. Qualtrics and Microsoft Excel v.14.5.5 were used for all analyses. Sample sizes varied by question since participants were allowed to skip any questions they did not wish to answer. Descriptive analyses about expectations for and use of WES results were prepared for the entire survey population.

Family Forum and Engagement

Partner-participants were invited to attend an annual family forum ("Duke Genomes Family Forum", DGFF) in years 2013-2015. Partner participants who did not provide contact information were excluded. Also, only partner participants for whom DNA sequencing had commenced (meaning all samples had been received) were included. Each attending family was offered \$100 to offset travel and/or childcare expenses for each event; most participant families were local/regional. On-site special-needs caregivers were provided to enable inclusion of

the children. The family forums were daylong conferences with activities, presentations, and social opportunities for families to meet one another and interact with attending researchers and clinicians. The agenda for each event included a mix of social events and presentations by researchers, clinicians, and participants. Participants attending the 2014 and 2015 DGFF events were invited to be interviewed for an independent documentary co-produced by study team members [25]. All partner participants were invited to a private screening of the documentary in early 2016. Participants attending the 2015 DGFF were invited to form a participant advisory board for the project.

Results

Participant Families

Between January 2012 and July 2016 (55 months), we screened 1,256 cases, ultimately enrolling a total of 225 families. Until June 2013, all families were enrolled on a genetics protocol (DUHS Pro00022846) that was not specific to exome or genome sequencing. The majority of participants thereafter enrolled on the more comprehensive exome and return-of-results protocol described herein (DUHS Pro00031066). Many of the participants consented originally on the former protocol were re-consented onto the exome protocol prior to return of results. Of the 225 consented families, 193 families (385 adults) were initially or eventually consented on the exome protocol. Self-reported race and ethnicity indicate that of the 450 adult parents (225 families), 106 (23.6%) parents were non-white, including 51 (11.2%) African or African American parents, while 51 (11.2%) parents had Hispanic ethnicity. Note that while 225 families enrolled in the study under the original protocol, not all families or family members re-consented to the exome protocol, leaving 385 adult consents on the exome protocol.

Consent Options

Of the 385 adults who enrolled on the exome protocol, 303 (78.7%) opted for "partnership" with the research team, while 82 (21.3%) opted for "traditional" participation. Of the 385 participants completing the exome consent form, four of the consent forms were blank for the options regarding return of direct results, indirect results, and future interest in non-clinical results, leaving 381 participants. No statistical significance was seen based on sex or race/ethnicity of participants. Nearly all adult participants (378/381; 99.2%) opted to receive results directly related to their children's conditions. The three who chose not to receive results were "traditional" participants. Most participants (353/381; 92.7%) also opted to receive additional clinically relevant results unrelated to their children's conditions. No statistical differences in choices were noted between partners who opted not to receive clinically relevant results and traditional participants. The majority of participants (315/381; 82.7%) also expressed an interest in receiving their own non-clinical results. Partners were somewhat more likely than traditional participants to elect this option, but this difference was not significant ($p=0.08$). No significant differences were observed between mothers versus fathers in selecting these

options; the numbers were too small to parse by other demographic criteria.

Survey Responses

The 303 partner participants were screened for eligibility for the survey. Excluded from this group were four participants pending consents or samples from one or both of the biological parents, and 32 participants who declined to provide email addresses. This left 267 eligible participants. Of these, 103 responded to the pre-exome survey (a 38.6% response rate) and 95 completed all questions in the survey (92.2% completion rate).

Of the respondents, the majority (80%) had a child under 10 with a condition that led to their interest in enrolling in the study. More mothers (78/103; 75.7%) responded to the survey than fathers (25/103; 24.3%; $p < 0.001$). Despite the relatively young ages of the children, almost a third ($n=103$; 29.6%:) reported that they had been seeking a diagnosis for their child for more than five years; 19.2% among those with children under 10 ($n=82$). In their searches for diagnoses, parents reported consultation with as few as one specialist (13/97; 13.4%) to more than 20 specialists (11/97; 11.3%). Overall respondents consulted an average of 6.7 specialists ($SD=6.19$).

On the scale responses, participants indicated that they expected to learn information about their children's conditions through the research study and showed a strong desire to participate in research (Figure 2). Participants also indicated that they had shared information about the research with both their family members and health care providers outside of the study team. Respondents indicated strong levels of trust in the study team and their children's doctors, but less trust of doctors not directly involved in the care of their children. Participants indicated that researchers had an obligation to tell participants about the genetic information they learned from the research and that

parents were entitled to access their children's genetic information. Respondents were split as to whether research made them nervous ($M=6.88$; $SD=2.78$) and if they were worried about the privacy of research data and samples ($M=5.88$; $SD=3.38$).

Survey respondents overwhelmingly indicated expectations for receiving genetic information related to the causes of their children's conditions, but also expected to get genetic information unrelated to their children's conditions, including susceptibility to adult diseases for themselves (81.2%) and their children (88.2%; Figure 3). All survey respondents indicated a desire to receive genome results pertaining to their children's conditions (Figure 4). Respondents also indicated a strong desire to know their own carrier status (96.9%) for autosomal recessive disorders, to receive their children's exome data (92.6%), and to receive their own exome data (85.3%).

Family Forum Participation and Project Engagement

Family forums were held during three consecutive summers from 2013-2015, with invitations extended to partner families. Each year a greater number of families qualified to attend as enrollment in the project increased. Invitations were sent to 70, 159, and 191 partner participants for each year, respectively. Absolute attendance remained fairly constant each year but declined proportionately as the overall number of participants grew: 21/70 (30%) in 2013; 27/159 (17%) in 2014; and 27/191 (14.1%) in 2015. Ten families were interviewed for the independent documentary; nine of the families were ultimately featured after one family declined continuing participation. The documentary was released to the public in 2017; the effect of the documentary on participants will be assessed in the future. In response to the study team's suggestion of developing a participant advisory board, two families briefly considered the possibility but no further steps have been taken outside of the study team's coordination.

Figure 2. Participants were asked to scale from 1-10, with 1 being “strongly agree” and 10 being “strongly disagree,” their (A) reasons for participating in the research project, plans to share research information, and (B) feelings about participating in research. The vertical line represents the mean (M) and the thick gray line the standard of deviation (SD).

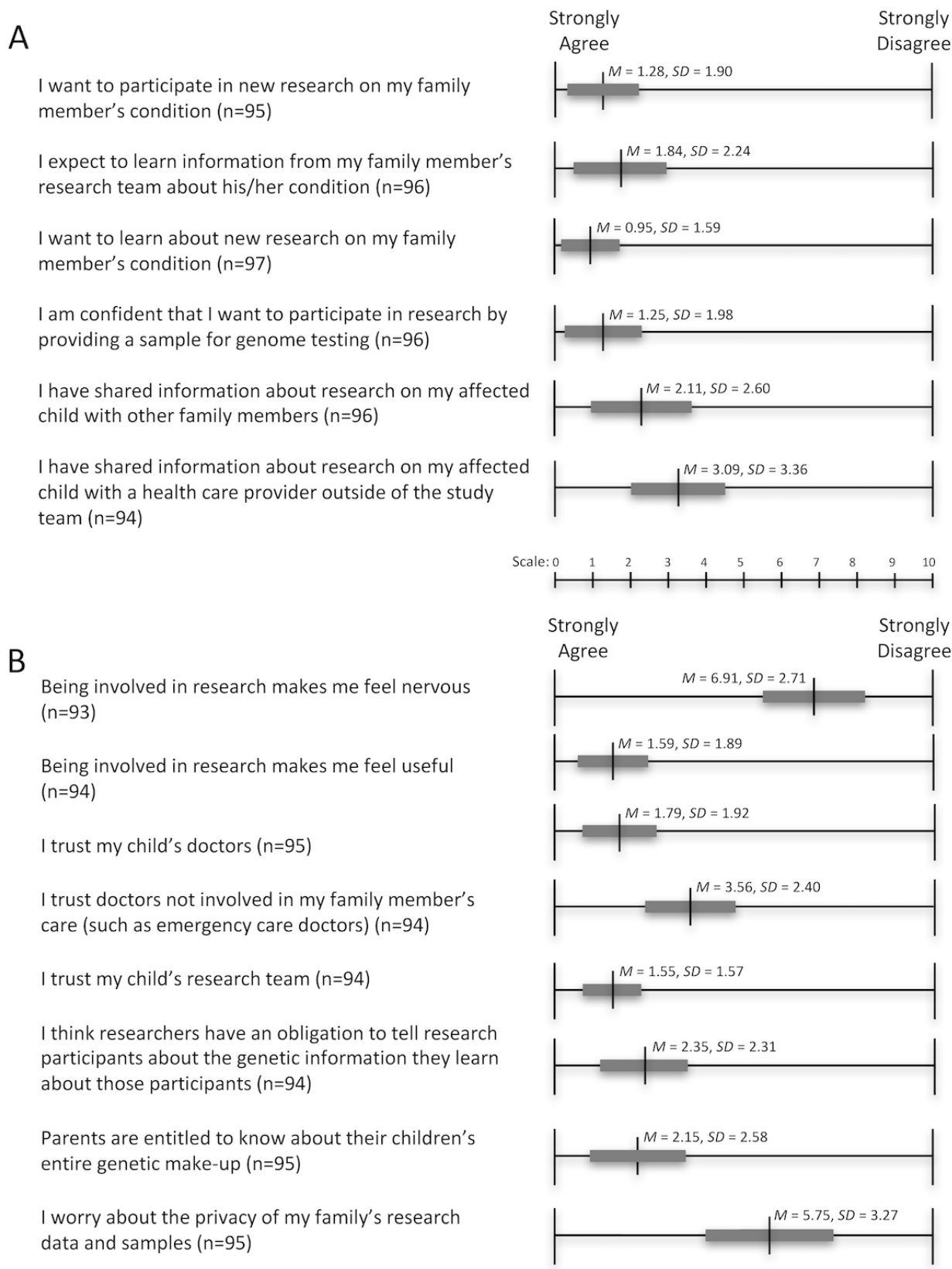


Figure 3. Participants were asked their expectations for receiving information (A) related to their child’s condition and (B) unrelated to their child’s condition.

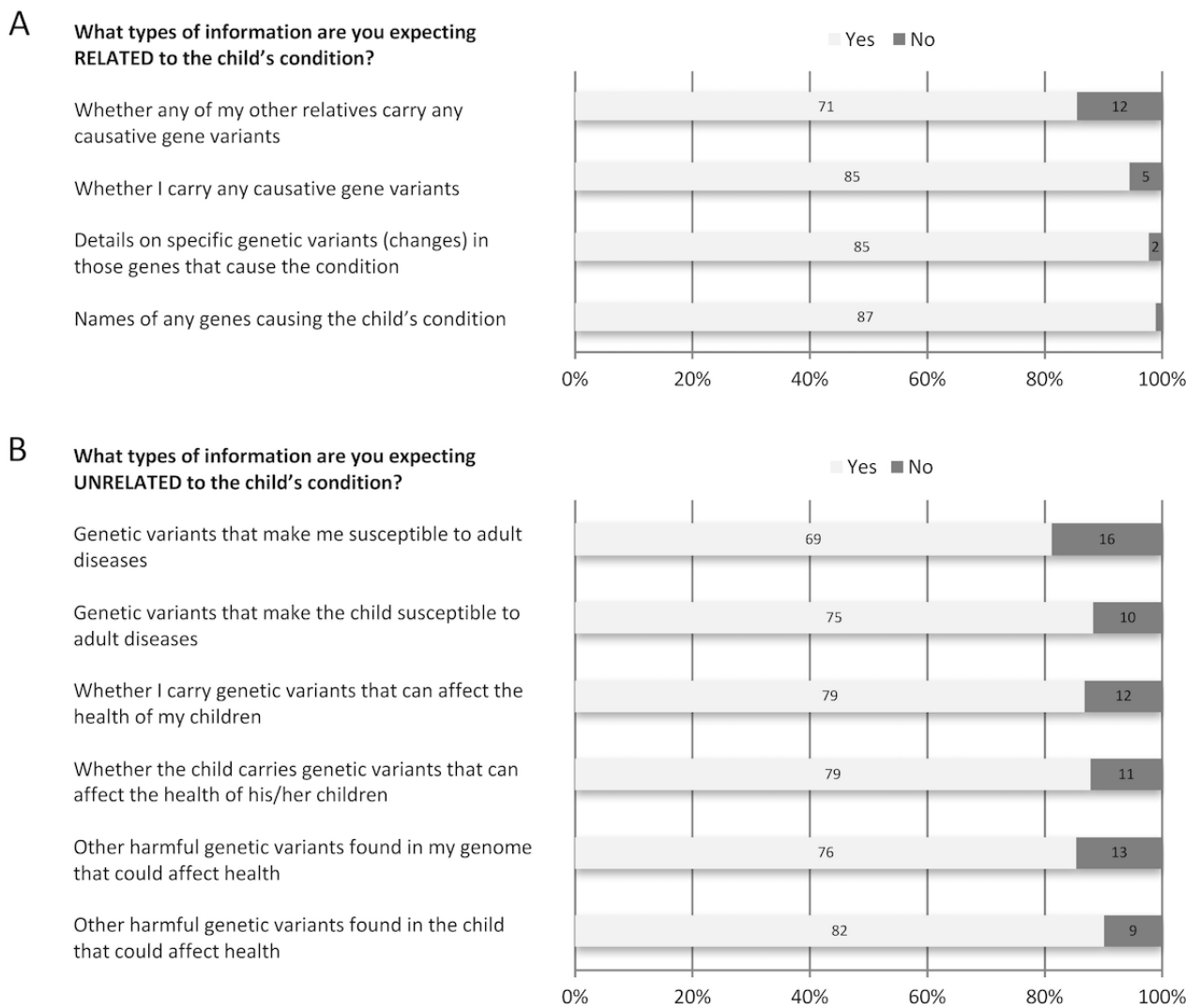
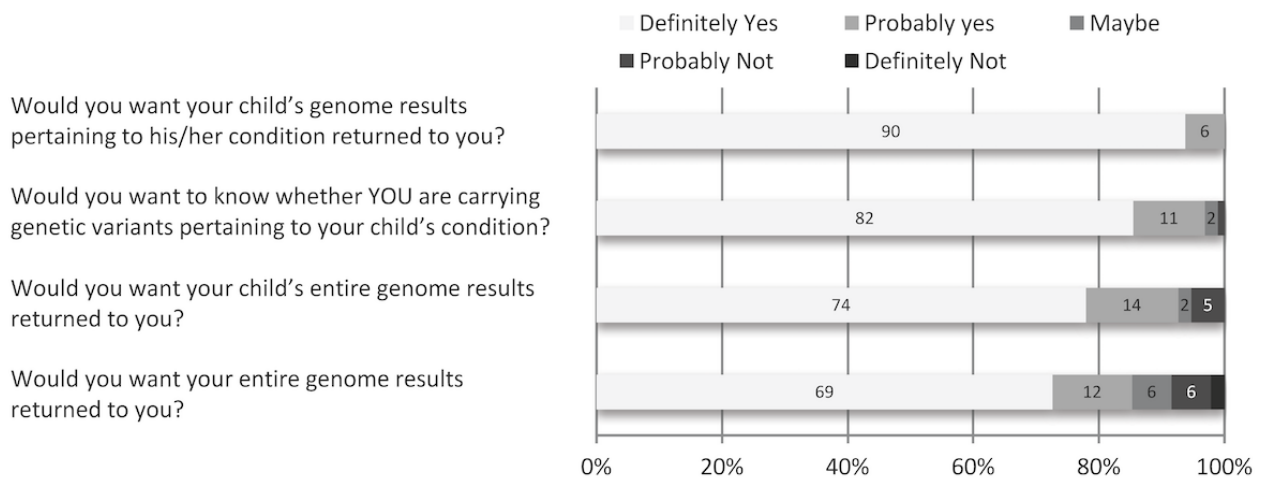


Figure 4. Participants were asked their desires for receiving information related to their child’s condition and for receiving their entire genome results.<



Discussion

We found that most adult research participants in a study of children with undiagnosed genetic diseases were strongly interested in obtaining genetic information about their children's conditions and about themselves. In this respect our findings are in line with previous work on return of results in families undergoing WES in search of diagnoses [26-29]. In addition, adult participants in the present study were generally highly trustful of the biomedical research enterprise, not anxious about research participation, and relatively unconcerned about the prospect of their privacy being compromised (Figure 2).

Presenting the participants with the option of "partnership" at the onset of the study enabled us to conduct the survey and to engage with participants via other opportunities such as an annual family forum and seminars. We could not assess the reasons to decline partnership, as our protocol precluded approaching traditional participants for additional research. As the partner families receive research results, we continue to invite them to Task Force events and to solicit their interest in ongoing survey research. The "traditional participant" option, which was chosen by 20% of the cohort, allowed participants to receive research results but to decline deeper engagement (and associated time commitments), including our surveys.

Families encounter numerous specialists and expert opinions and may be enrolled in numerous research studies. Most families in our study have been seeking a diagnosis since the birth of their child; 11% had seen more than 20 specialists. Until a genetic etiology is determined, they often have few opportunities to engage with other families experiencing similar social and medical challenges. The overarching objective of our research study is to understand human genetic variation on a broad scale; but of course, this is not mutually exclusive with individual families learning something from their unique situations and their own particular genetic variants. We have tried to offer families opportunities through the family forums and other gatherings to learn about how the research is done, why it can take a long time, to meet other families seeking a genetic etiology for their children's medical conditions, and to provide feedback to researchers and clinicians on their experiences and expectations. We co-produced an independent documentary film that followed families' experiences in seeking diagnoses, enrolling/participating in our study, and negotiating the daily challenges of living with special-needs children.

We see partnership -- that is, open exchange of genetic research information and shared decision making -- as the most equitable framework for large-scale genomic studies and the one with the highest upside for researchers, clinicians and patients [30]. However, partnership comes at a high price and with significant challenges. The effort of a research team to engage individually with participants is significant, both in expense and time. The long turnaround time associated with genome sequencing, analysis, and modeling specific variants in zebrafish limits the pace with which we have been able to return final results, which frustrates clinicians, participant families and researchers alike. And obviously, families raising children with special needs have priorities that start with care of their children; engagement

in research beyond provision of a genetic sample is likely to be of interest only when they believe they may realize some clinical and/or personal benefits [31]. Many families were unresponsive to invitations to the family forums, even with a small financial incentive for participation (though it is possible the incentive was insufficient). The families attending the family forums reported satisfaction with their experience. Eight participants attending the 2015 forum completed a post-family forum satisfaction questionnaire and no significant negative experiences were reported. For example, one participant commented:

It was really nice to be able to meet other families who are part of the study. It makes the study feel like it is more than just a study with 'subjects.' We also enjoyed being able to learn more about the study/research beyond our family.

However, the opportunity to meet other families negotiating similar circumstances, ongoing leadership among partner families for future community efforts has not coalesced. Attendance at the annual forums did not grow with the pace of the program and families have not come together to develop a participant advisory board that might influence the direction of the research and the institution's approach to families with undiagnosed children.

Moreover, while the survey indicated a strong desire for receiving personal genetic data, at our institution the mechanisms and policies to enable research data sharing with participants continue to lag behind some other initiatives (eg, Geisinger; MyGene2). The inability to meet participant expectations can create frustration among participants who want information and among researchers who are reticent to provide incomplete and potentially uncertain data.

There were several limitations to our study. They include nonrandom ascertainment, ie, referring physicians were apt to be part of the Duke Health System and thus known to and trusted by participating families. In addition, given the rhetoric of partnership present in recruitment and online materials, our sample may well have been subject to a self-selection bias. Moreover, we did not include families who read the consent but ultimately chose not to enroll; their decision could easily have been influenced by privacy concerns—clearly this is a subject deserving of attention in subsequent research.

Diagnostic exome sequencing is less than a decade old. Remarkable progress has been made and the reference databases and number of sequenced exomes have grown exponentially [32-34]. At the same time, survey data have made it clear that genomic research participants expect to receive individual results [7,10,35,36]. Meanwhile, participants in the emerging biorights movement are refusing to contribute samples without assurances that they will: (1) be financially compensated; (2) receive relevant individual medical research information; and/or (3) be able to exercise some measure of control over the fate of their samples and data [37]. Moreover, the US Department of Health and Human Services Office for Civil Rights' interpretation of the recent changes to CLIA and HIPAA suggests that research participants whose sequencing/genotyping was done in a HIPAA-covered lab have broad access rights not only to final

interpreted test reports, but to all underlying genomic data that is traceable to them [38,39]. Thus, the partnership turn is now not only a moral and popular one, but a legal one.

That said, the ways in which we negotiate data access and participant expectations are a work in progress. Our hope is that the frank and forward-looking commitment of the National Institutes of Health to share individual results with large

numbers of participants [40] will lead to: (1) the construction of a robust infrastructure for sharing genomic data and engaging with research participants; (2) active formation of support networks among other families living similar experiences involving genetic disease and uncertainty; and (3) a pervasive change in culture, that is, a day when information asymmetry is supplanted by true partnership.

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Conflicts of Interest

None declared.

Authors' Contributions

The authors contributed directly to this article. In addition, this work was facilitated by the Duke Task Force for Neonatal Genomics consortium that manages the research of undiagnosed pediatric conditions presented at Duke clinics. The consortium is comprised of a Clinical Ascertainment team (June Allison-Thacker, Alexander Allori, Susan Anderson, Patricia Ashley, Margarita Bidegain, Brita Boyd, Eileen Chambers, Heidi Cope, C Michael Cotten, Sarah Ellestad, Kimberley Fisher, Joanne Finkle, John Foreman, Amanda French, William Gallentine, Rasheed Gbadegesin, Ronald Goldberg, Kevin Hill, Pearce Jackson, Sujay Kansagra, Vellore Kasturi, Joanne Kurtzberg, Monica Lemmon, Amanda Marion, Ann Mabie-Deruyte, Jeffrey Marcus, Marie McDonald, Stephen Miller, Mohammed Mikati, Amy Murtha, N Christine Øien, Kristin Page, Yezmin Perilla, Carolyn Pizoli, Todd Purves, Sherry Ross, Azita Sadeghpour, Edward Smith, Jennifer Stout, Jessica Sun, John Wiener), a Genomic and Functional Analysis team (Shanté Adams, Erica E Davis, Shannon Dugan-Rocha, Ludmilla Francescato, Richard A Gibbs, Christelle Golzio, Carter Gunn, Nicholas Katsanis, Tahir Khan, Amalia Kondyles, Maria Kousi, Zachary Kupchinsky, Kristin McDonald, Kelsey McFadden, Natalie Mola, Donna Muzny, Adrienne R Niederriter, Aysegul Ozanturk, Igor Padiaditakis, Kavita Praveen, Ellen Richardson, Holly R Robertson, Chad Russell, Jeremiah Savage, Aniko Sabo, Karen Soldano, Vanessa Thomas, Jason R Willer), and an Ethical, Legal, and Social Implications team (Misha Angrist, Robert Cook-Deegan, Sara H Katsanis, Mollie A Minear, Ashley Warman).

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Abbreviations

- CLIA:** Clinical Laboratory Improvement Amendments
DGFF: Duke Genomes Family Forum
HIPAA: Health Insurance Portability and Accountability Act
PMI: Precision Medicine Initiative
SD: standard deviation
TFNG: Task Force for Neonatal Genomics
WES: whole exome sequencing

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Original Paper

Patient Experiences in a Linguistically Diverse Safety Net Primary Care Setting: Qualitative Study

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Abstract

Background: The patient-centered medical home model intends to improve patient experience and primary care quality. Within an urban safety net setting in Northern California, United States, these desired outcomes are complicated by both the diversity of the patient community and the care continuity implications of a residency program.

Objective: The objective of our study was to understand the patient experience beyond standardized satisfaction measures.

Methods: We conducted a qualitative study, interviewing 19 patients from the clinic (English-, Spanish-, or Mien-speaking patients).

Results: Some themes, such as the desire to feel confident in their doctor, emerged across language groups, pointing to institutional challenges. Other themes, such as distrust in care being provided, were tied distinctly to speaking a language different from one's provider. Still other themes, such as a sense of powerlessness, were related to cultural differences and to speaking a language (Mien) not spoken by staff.

Conclusions: Findings illuminate the need to understand cultural behaviors and interactional styles in a diverse patient population to create a high-quality medical home.

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KEYWORDS

safety-net providers; urban health services; primary health care; patient-centered care; qualitative research; quality of health care; communication barriers

Introduction

I don't know because I don't understand English, and then whatever I tell the interpreter, he is relaying the information in English, and then relaying what the doctor says back to me. There is a gap.
[Mien-speaking patient]

This comment was shared by a Mien-speaking patient at the Lakeview Hospital Adult Medicine Clinic (LHAMC, located in an urban setting in Northern California, USA; we have changed the name of the hospital to protect the privacy of the patients) and provides a window into one of the challenges experienced by non-English-speaking patients within a multilingual, multicultural urban safety net setting.

Patient-centered care is an increasingly promoted approach for improving quality of, access to, and satisfaction with health care, particularly for primary care [1,2]. To fully implement patient-centered care, we must partner with patients and understand how to strengthen their experiences and outcomes [3]. Many factors shape the way patients perceive quality of care. In urban safety net clinics, understanding those experiences can be challenging. Different constructs of health, levels of acculturation, and institutional barriers implore clinicians to find new methods to explore patient experiences [4]. When patient views are solicited through qualitative methods, results emphasize aspects of the patient experience that may have otherwise gone unnoticed and could effectively shape patient experience, clinical functioning, and patient outcomes [5].

At the time of this study, the LHAMC was beginning the conversion to a patient-centered medical home (PCMH) model of care. The proposed operational changes required that clinic leadership better understand the views of patients in order to incorporate specific interventions that would increase patient satisfaction. The conversion also brought the opportunity to explore some of the barriers to patient satisfaction, such as the transient nature of resident physicians providing primary care and language barriers faced by the patients, who speak over 35 different languages.

The presence of resident physicians may contribute to fragmented care and other negative patient experiences that compound the challenge of providing high-quality patient-centered care. At the time of this study, approximately 50% of the clinic's patients received their care from resident physicians in an internal medicine training program that used the traditional curricular model of a weekly continuity session during all rotations. Residents can positively influence quality of care and outcomes by developing close interpersonal relationships with patients [6]. Conversely, when a resident cares for patients with low English proficiency, misuse or underuse of interpreter services by the resident may negatively affect the patient's perception of quality of care [7].

Language barriers can also contribute significantly to differences in patient satisfaction. Patient-provider language discordance may negatively influence patient experience, leading to the patient disclosing less information and feeling negatively judged, vulnerable, disrespected, and helpless [8]. Furthermore, in language-discordant provider-patient interactions, time constraints, availability of interpreter services, and other institutional challenges can lead to disparities in the quality of the communication experience, patient satisfaction, and patient health outcomes [9-12]. To help understand the impact of language on the patient experience at LHAMC, we applied the concept of *cultural health capital* (CHC). The CHC framework "provides a way to understand how features of patient-provider interactions—such as interpersonal rapport, exchange of information, empathy, and trust—are accomplished or undone, based upon the repertoire of specialized cultural resources that patients bring to the health care encounter, in combination with providers' fostering of and receptiveness to those resources" [13]. This theoretical model was important in analyzing the clinical interactions described by the LHAMC's patients.

The Building Together Project began in January 2014. As the LHAMC continued its process of converting to a PCMH, staff and researchers recognized the need to engage directly with members of the urban safety net clinic's diverse patient community. A team of 1 attending physician (LB), 1 resident, 2 medical interpreters, 1 researcher (RLB), and 1 English-speaking patient came together with the goal to use qualitative research methods to explore the following research questions: (1) What makes a clinic experience for a patient positive, and why? (2) What makes a clinic experience for a patient negative, and why? (3) How do patients' personal, cultural, and historic contexts affect their clinic experience? By gaining a deeper understanding of diverse patient experiences within the LHAMC, we hoped to provide a more nuanced foundation on which to build ongoing patient-centered quality improvement.

Methods

Setting

Lakeview Hospital is part of a county health system and sees over 180,000 outpatient visits a year, serving patients who speak over 35 languages. The LHAMC is a primary care medical home for adult patients within the hospital, seeing approximately 8000 patients a year for preventive, acute, and chronic care. Over 80% of patients use Medicaid, and 15% use the county health program for uninsured residents.

We focused on 3 patient language groups: English-speaking, Spanish-speaking, and Mien-speaking patients. English and Spanish were the 2 most commonly spoken languages within the LHAMC, listed as the primary language spoken for 73% and 18% of patients, respectively, in fiscal year 2013. Speakers of Mien, who originate from southern China and northern Southeast Asia, and who made the largest percentage of recorded LHAMC interpreter requests in fiscal year 2013 (29% of requests), are not often able to participate in English- or Spanish-language feedback opportunities. Mien is a traditionally oral language, which could lead to unique challenges in navigating the hospital system.

The Building Together Project study procedures were approved by the health system's institutional review board (IRB14-02041A).

Interview Guide

To ensure that the interview guides and protocol reflected the experiences and perspectives of the diverse members of the team, the team's researcher member facilitated an experiential training on qualitative research and interview guide development with team members. After discussing the subjective paradigm and text-based data of qualitative research [14], the team reviewed the structure and function of an interview guide and reflected on the 3 driving research questions of the project. The team then developed our own clinic patient experience wheel to identify the aspects of the patient's experience that they wanted to ensure could be explored, based on an example from Western Australia [15]. The wheel shows the different experiences that patients move through as a part of a visit to the clinic, beginning with getting to the clinic and concluding with

discharge and follow-up for future appointments, which leads back to the first step (see [Figure 1](#)).

After creating the wheel, the team identified 3 overarching aspects of patient experience: (1) space and procedures of the clinic, (2) relationships with staff in the clinic, and (3) experience of care and beliefs about health. These 3 aspects became the topical frames for the study’s interview guide. The team composed questions, ensuring that the language would translate well from English into Spanish and Mien.

Interview

Patients were recruited in the waiting room of the LHAMC. The researcher along with the team’s interpreters conducted face-to-face interviews in a room nearby but outside of the clinic so as not to conflate the interview experience with the patient’s clinic visit. We set the goal of interviewing 16 to 24 patients (8-10 English-language, 4-6 Spanish-language, and 4-6 Mien-language interviews) based on resource and time constraints, as well as the hope of achieving thematic saturation related to language- and culture-specific issues within each group [16].

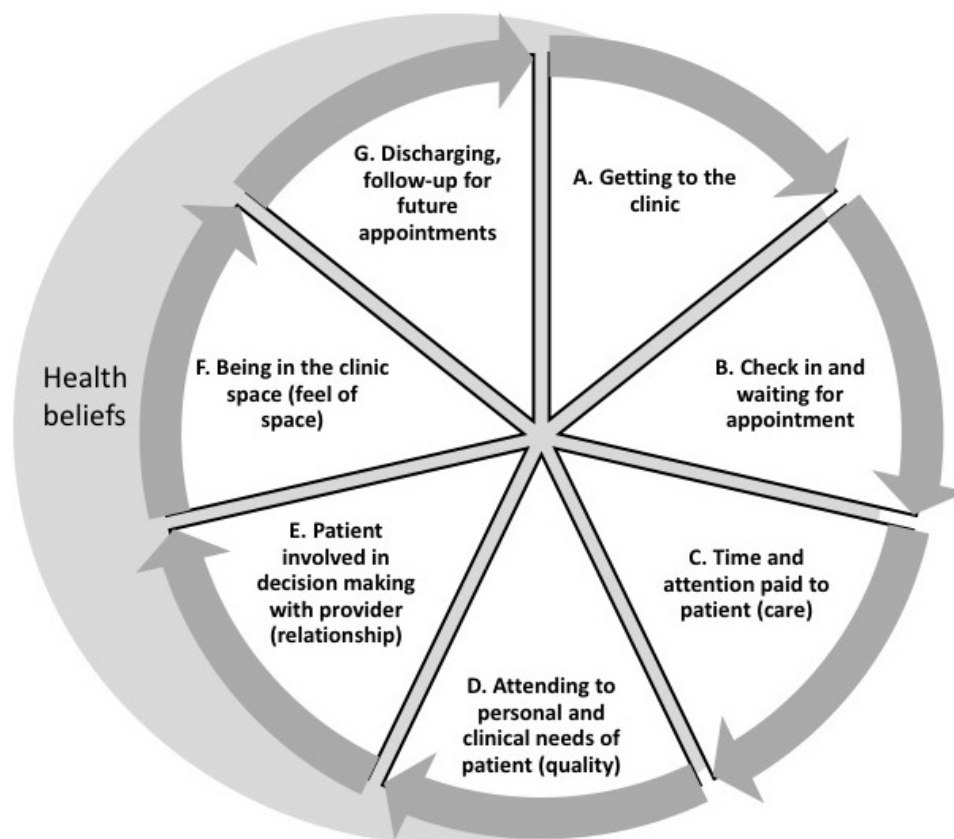
All interviews were conducted in English (with support from a Spanish-speaking or a Mien-speaking interpreter for those language-specific interviews). After each interview, the interviewer and interpreter had a debriefing conversation, and

the interviewer recorded notes if shifts or changes were made to the interview guide or procedure. Following the first interviews in each language group, the team made additional minor changes to the structure and wording of questions. Most notably, the team shifted the Mien-speaking patient interview guide to accommodate the refusal of Mien-speaking patients to have their voice recorded, either for fear of ramifications or a belief that recording one’s voice could ultimately trap one’s soul. Accordingly, the Mien-speaking patient interview guide provided space for the researcher to write notes to record Mien-speaking patient responses to questions and probes. Patients received a US \$20 gift card for completing the interview.

Analysis

This study used thematic analysis, a methodology that manifests differently depending on specific parameters set by a study’s researchers, emphasizing the importance of transparency on the part of researchers in articulating the assumptions, decisions, and actions that lead to the ultimate analysis of themes [17]. Of priority was incorporating the team’s diverse perspectives into the development of codes and analysis of transcripts, drawing on the principles of community-based participatory research, in which the inclusion of nonacademic researchers connected to the circumstances of interest throughout the research process enhances the investigation and findings [18].

Figure 1. Patient experience wheel for a multilingual, multicultural urban safety net clinic.



The recorded interviews from the Spanish-speaking and English-speaking patients were transcribed throughout data collection by the researcher and a transcription assistant. Notes from Mien-speaking patient interviews were typed up by the researcher as soon as possible following the interview to capture as many of the Mien-speaking patient's own words as possible.

Coding

The team used an adapted consensus-building approach with a sample of 6 representative transcripts (2 from each language group) to develop a codebook that could be applied to the remaining transcripts. First, all participating team members applied the coding steps to the same part of one transcript. The group compared findings, looking for similarities and differences, ultimately coming to an agreement about the overarching codes. Next, 2 pairs of 2 team members were given 2 transcripts or notes from patients of different language groups to code. Within the pairs, each individual completed the coding steps independently and then discussed the identified codes with his or her coding partner, ultimately coming to a consensus around the codes present within the transcript (or notes).

Codes and definitions were sorted and grouped into similar categories. This process resulted in 8 overarching codes: (1) continuity of care, switching doctors, and resident turnover; (2) health care system, structure, and navigation; (3) relationship and communication between staff, health care team, and patient; (4) access to and experience with interpreters; (5) language and culture differences or similarities; (6) experience of waiting; (7) what it means to be healthy or unhealthy; (8) perspectives on needed improvements in the clinic.

The researcher, attending physician, and patient next independently coded the remaining transcripts using this codebook. Finally, the researcher reviewed all coded transcripts and notes to ensure that all aspects of the interviews relating to the 8 codes were captured.

Thematic Analysis

The researcher and attending physician reviewed coded segments within specific codes to identify themes relevant to answer the research questions. The team reviewed these themes to ground the findings in the team members' unique experiences and perspectives. The researcher then looked across all codes, comparing ideas within and between language groups to identify broader patterns of significance in relation to the initial research questions.

Results

Patient Demographics

Between February and May 2014, the team conducted 19 interviews—8 in-person and 11 over the telephone—with recordings ranging from 13 to 60 minutes. [Table 1](#) presents the demographic characteristics of the participants. English-speaking patient participants were younger, more likely to be born in the United States, and more likely to have some education than were the Spanish-speaking and Mien-speaking patients.

Common Themes Across all 3 Language Groups

A Personal Relationship With a Provider—Feeling Cared For

Patients from all 3 language groups described what made a good doctor-patient or staff-patient relationship, including the desire to feel confident in their doctor, and to have their doctor listen to them and engage with them, explain treatments and options, and show concern for their well-being.

[An ideal interaction with a doctor] would be one on one. And also that the doctor or practitioner would engage me too. To see you in the eye to see you as a person and not just a statistic. [English-speaking patient]

Table 1. Interview patient demographics.

Characteristics	All patients (N=19)	English-speaking patients (n=8)	Mien-speaking patients (n=6)	Spanish-speaking patients (n=5)
Age (years), mean	55	46	67	56
Country of birth, n				
United States	7	7		
Mexico	6	1		5
Laos	5		5	
China	1		1	
Education, n				
Some	14	8	2	4
None	5		4	1
Work, n				
Not working	16	5	6	5
Working	3	3		
Female sex ^a , n	11	5	3	3

^aSex was defined by the interviewer and not by the patient.

“Knowing My History”

Within reflections on relationship, patients emphasized the importance of their doctor knowing their history, and the frustration experienced when a doctor asked repetitive questions or clearly suggested in other ways that the doctor was unaware of the patient’s history, wasting the limited time of the visit.

I feel like it’s not right for me to start talking again, and again and again every time I go to see a doctor and every time I been seen by someone new to start talking about my old medical issues, my old problems. So I want them to focus on what I am telling them at the moment but they just keep asking me question about the old stuff. [Spanish-speaking patient]

The Challenge of the Resident Doctor

Patients discussed the impact of having a resident as their doctor, including feeling that the doctor was just using them for training and would abandon them, feeling confused as to why doctors left after their training, or feeling as if their doctor was not a “real” doctor yet.

I believe that they are not a real doctor...I feel like they just use me as an object for their training...If he is my doctor, he is always there to help me, he knows what I need, he is there to help me. But with the residents, it’s like a girlfriend and boyfriend—if I like you, I’ll stay for a little. If I don’t I’ll leave...if they don’t like caring for you, they go away. [Mien-speaking patient]

Communication Issues Within Lakeview System

Patients described challenges with understanding what and why changes were happening at the clinic level (such as a delay getting an appointment), and with their ability to speak directly with doctors about issues. Patients also described challenges with the LHAMC communicating with other clinics within the hospital system to coordinate care or to follow up on tests or treatments needed.

Experiences varied, however, as some patients offered examples of good-quality communication experiences.

Well, I from the very beginning, I notice even when I start getting to the desk information. I notice that the information given to me was very clear...at Lakeview there are signs where you need to go to ask for information and that makes the, um, services to be better, more effective, and what you need to do. [Spanish-speaking patient]

Waiting

Across language groups, patients discussed the frustration with waiting to be seen during their appointment at the clinic, waiting to get an appointment, or waiting in other contexts (including the pharmacy) within the hospital.

[I]t’s just very its frustrating because they are so slow its just so slow it takes long times to be called up there to register and then you have to finally get registered which is gonna be after your appointment and even though I come early it takes them so long to register

to call you up to register you that they call you after your appointment. But it doesn’t matter because the doctor is gonna take forever (laughing) to see you so umm it’s just really frustrating that’s what I hate about Lakeview the waiting the slowness that and that’s for the whole hospital for everything. Everything is just slow. [English-speaking patient]

Themes Related to the Added Impact of Language

The Mien-speaking and Spanish-speaking patients highlighted experiences and realities, not described by the English-speaking patients, that were particular to the realities of speaking a language other than English within the health care system. Within these themes, the experiences of Mien-speaking and Spanish-speaking patients also differed based on the commonality of each language among clinic staff.

Impact of Language on Relationships

Mien-speaking and Spanish-speaking patients emphasized the added impact of language differences and commonalities on establishing trust and a good relationship between patient and provider and feeling confident that the patient’s needs were being understood.

Maybe I don’t speak English, they treat me differently. I look at their actions. Maybe with English-speaking patients they change and act differently. [Mien-speaking patient]

[Seeing a doctor who does not speak Spanish] makes the visit to the doctor’s more difficult because we don’t have any clear communication. Maybe the symptoms will be, uhh, not interpreted correctly. [Spanish-speaking patient]

Some Mien-speaking patients also suggested that not speaking the same language as clinic staff or providers could have an impact on their overarching experience in moving through the appointment process.

Waiting [in the waiting area] is also hard because I have to wait a long time, sometimes a half hour, sometimes more, one and a half hours. For me, I don’t have to wait long but my mom sometimes waits all day. I suspect because she does not speak English and cannot push at registration. [Mien-speaking patient]

Mien-speaking and Spanish-speaking patients also talked about other ways in which providers reached across language barriers to establish trusting relationships, including an emphasis on tone and touch (Mien), trying to speak some Spanish or identifying a new provider for a patient who did speak the same language (Spanish), and insisting to the interpreter that all of the information that the patient wanted to share was important to the doctor (Spanish).

[My] doctor is good. She understands, the way she talks is very caring. She is not like other doctors, other doctors don’t have time to listen to you, and then their tone of voice is harsh. She understands culturally and she takes time to listen to a patient. [Mien-speaking patient]

And he was always very kind, always trying to help me but every time I saw him, that was with the help of an interpreter. And I think that he understood a lot because sometimes when I was telling the interpreter there's some things the interpreter used to tell me, "Well that's not important for the doctor to know that" or "that is not something that the doctor can help you with that," so and I was just telling the interpreter and many times the doctor say, "What, what they are telling me? No, no, tell me. What is what she wants? Tell me what is what she saying?" And the interpreter used to say, "Well it's not something that is important for you, doctor. That is an area different to yours." And he always say, "No, I wanna know. I want to know anything related to the patient." [Spanish-speaking patient]

Role of the Interpreter

Mien-speaking and Spanish-speaking patients discussed the role of the interpreter as a facilitator of conversation and care between doctor and patient. While many interpreter interactions described by patients referred to in-person interpretation, some patients did describe the use of the video interpreting system. Most of these patients found that experience to be positive, with one Mien-speaking patient stating explicitly that face-to-face interpretation was preferred. Most patients saw having an interpreter as a positive and vital part of the relationship (and insisting on having an interpreter present was seen as key).

My problem is that I don't speak English, but when I see a doctor, there are interpreters that help me through the machine, and so that is good, that is very helpful for me. And I don't have any problem with that. And what I see is that everybody who goes there, there is no distinction, there is no discrimination, everybody is being seen in the same way and treated in the same way, so that is good for me. [Spanish-speaking patient]

I would like to see somebody who pays attention to me, who can hear my concerns, who can communicate with me better, who don't assume that I speak English. My medical terminology is not so good, so get an interpreter for me. [Mien-speaking patient]

Other patients emphasized the potential for things to be lost in the conversation or not feeling confident that the interpreter was adequately conveying the needs of the patient, which was detrimental to the relationship. The power held by the interpreter as the arbiter of conversation was highlighted as potentially problematic.

Waiting to Speak

Spanish-speaking and Mien-speaking patients discussed the additional frustration of waiting just to speak—waiting to have an interpreter available (in person or through the video interpreting system) and then waiting during the visit itself while the conversation was being translated back and forth. Waiting for an interpreter was particularly frustrating, given how little time patients felt they had with the doctor during an

appointment. Patients also discussed this adding to a sense of feeling bad because they did not speak English.

[S]ometimes there is a little problem because the nurses do not speak Spanish...and sometimes they need to call or bring someone who speaks the language and sometimes they cannot find it... [so] they try to look for someone to come in an interpreter, or they can, they look for someone who is nearby next to me who can speak Spanish so they use the person... [and] it makes me feel a little bit bad because I do not understand English. [Spanish-speaking patient]

[Explaining why it would be better to just use a family member as an interpreter] The reason is if it's not difficult, then you can use the family member, hurry, finish up, and go home. Because the interpreter may be busy and you may have to wait longer. Yeah, wait a very long time, sometimes they have to finish what they are doing. [Mien-speaking patient]

Discussion

Principal Findings

Across the 3 language groups, patients emphasized the importance of a good-quality relationship with their doctor and staff. They highlighted the importance of empathetic listening, supportive explanations of health issues and treatments, and a demonstration of understanding a patient's history during a visit. Qualitative and quantitative studies alike emphasize the importance to patients of clear and positive communication with providers (and ancillary staff) in which providers and staff listen to their patients, show concern for their well-being, and spend time to clearly explain health issues, treatment options, and other procedural realities [3,8,19].

Patients described the unique challenges of having a resident as one's primary care physician and engaging with a residency training program overall, emphasizing in particular the challenges with abrupt and at times unexplained discontinuity with resident providers, as well as a negative feeling of being "trained on" and not being seen by a "real" doctor. Studies provide evidence of the importance of provider continuity for patient satisfaction, reduced emergency medical use, and even some health outcomes [5,20]. Traditional residency training schedules can make it difficult for patients to achieve a sense of interpersonal or relational continuity (terms that describe long-term, trust-based relationships between providers and patients in which patients sense a provider's commitment to the patient's well-being) with their resident physicians [21]. In addition, residents receive limited training in the use of professional interpreter services and may underuse these resources, particularly if they have some proficiency in a patient's language or if a patient brings a lay interpreter such as a family member to a visit [22-24]. Since this study, LHAMC's residency program was converted to a 3+1 curriculum in which residents spend 1 out of 4 weeks in the clinic without interference from any other clinical responsibilities. In addition, LHAMC has formed a team system in which each resident works closely with faculty, nurse practitioners, and nursing staff, who provide continuity during

the 3 weeks when the resident is on another service. These changes appear to have resulted in improved patient satisfaction with resident physicians as continuity providers as measured by a recent survey of 200 sample patients. As measured by the annual US Accreditation Council for Graduate Medical Education survey, the residents have voiced a significant increase in their satisfaction with practicing primary care in the clinic.

For the Spanish-speaking and Mien-speaking patients, speaking a language other than English added another layer of complexity and difficulty regarding basic interaction with doctors and staff, as well as interaction with residents specifically. Patients described a general concern as to whether doctors and patients fully understood each other when having to work through an interpreter. Patients highlighted that doctors and staff did try to reach across the language barrier to establish positive relationships. For Spanish-speaking patients, however, identifying a Spanish-speaking provider was seen as ideal. For Mien-speaking patients, the option of identifying a language-concordant provider or staff members is not yet possible within the LHAMC, and this reality can lead to feeling lost in spaces such as the waiting room. Mien-speaking and Spanish-speaking patients alike emphasized the importance of having an interpreter available when language concordance with a provider is not an option. Patients also highlighted the frustration of waiting for an interpreter, waiting just to speak, particularly when appointment times are so truncated. Mien-speaking patients described wishing a family member could be used to speed up the waiting time. While the potential benefits of having a strong advocate for the patient serve as the interpreter are important, concerns regarding the accuracy of information being transferred are also important to consider [25,26].

Studies have found that having an interpreter can add time to the length of a patient visit, particularly in relation to the interaction with the provider [27]. While video- and telephone-based interpreting systems may decrease waiting times for interpretation, remote interpretation systems may not decrease the length of the visit itself or may not be preferred in relation to the visit quality [27,28]. In surveys, LHAMC physicians have also voiced the concern that patients who are not language concordant should be given the same amount of appointment time as patients who are language concordant with their physician. This either limits the quality of the visit or results in longer visits, increasing wait times for subsequent patients.

The added impact of a language difference on the patient experience of primary care, which is tied to feeling confident in developing a good-quality relationship with a provider, can be understood in the context of CHC [13]. CHC encompasses the various skills, cultural understandings, and attitudes that allow a patient to satisfactorily navigate the health care system and patient-provider interactions. Through the lens of CHC, lacking such competencies creates and perpetuates inequities within a health care setting.

Patient characteristics constituting CHC include “knowledge of medical topics and vocabulary,” as well as “the skills to

communicate health-related information to providers” [13]. While difficulties with these characteristics are not limited to non-English-language speakers, the situations described by Mien-speaking and Spanish-speaking patients—having to either communicate in a second language or communicate through an intermediary—add another barrier to learning and using CHC to achieve better quality of care. All of the English-speaking patients had some education, and most were born in the United States, potentially enhancing the ability of these patients to navigate the predominantly English-speaking US health care system. As one Mien-speaking patient discussed, not speaking English also hindered self-efficacy related to engaging with the registration staff to understand the delay being experienced by the patient’s mother. This touches on another component of CHC—having “an enterprising disposition and a proactive stance toward health” and one’s care [13]. While CHC is something that one can cultivate over time through repeated interaction with providers and the health care system, when one does not speak the same language, it may be more difficult to gain a full embodiment of these characteristics. If there is a fundamental uncertainty as to whether a given encounter is being fully understood by a provider, how can a patient build the habits and instincts that can enhance the patient experience and quality of receiving care?

CHC is not limited to identifying the characteristics that patients need to effectively maneuver through a health care interaction. The CHC concept also emphasizes the interpretation of CHC characteristics by physicians, stating that the physician’s interpretation can affect how he or she unconsciously perceives and ultimately treats the patient. As Shim describes, “patients and family members who mobilize CHC to present themselves and their health issues in approval-garnering and medically intelligible ways can generate ‘cascades’ of subsequent interactions and actions...that may enhance communication and care” [13]. The opposite is also true. If a physician notices a patient’s lack of CHC characteristics—such as the ability to communicate effectively about medical circumstances—the physician may inadvertently alter the way that he or she provides care, giving an impression of impatience or lack of concern akin to what some patient participants in this study noted. As the patient participants described, feeling as if a doctor is not actively cultivating the patient-provider connection can have a detrimental effect on the patient experience.

Limitations

Limitations of interview structure and time precluded our abilities to explore cultural perceptions of health and well-being in depth, which would have added to the understanding of diverse patient communities’ engagement with primary care, as well as the broader application of the CHC structure. Spanish-language and Mien-language interviews were conducted with support from interpreters; while the researcher and interpreters took care to ensure clear communication throughout the interview experience, there remains a potential for some information to have been lost or misconstrued in translation. Also important to note, the study could not encompass the full ethnic and linguistic diversity of the Spanish-speaking and English-speaking populations of LHAMC in the 13 interviews conducted for this study. Indeed, engaging with the full linguistic

and ethnic diversity of the clinic was beyond the scope of this study. While themes were repeated within each language group, suggesting a degree of saturation in some thematic areas, additional interviews may have revealed further elements of the patient experience.

Implications

The process of recruiting patients and conducting the interviews for this study was the basis for important patient-centered quality improvement efforts within LHAMC. Projects to date have specifically addressed the themes that emerged from these interviews. For example, having access to a provider who knows the patient's medical history was addressed by dividing the staff into 4 care teams who cover for one another and share information routinely. Since this study, the clinic has adopted an electronic health record system and have implemented standard operating procedures, which require a previsit medical record review and huddle with the care team. Patient flow has improved and a waiting room protocol developed by the council was introduced to inform patients of the estimated wait times.

In addition, building on the lessons learned from the patient interviews, the clinic received external funding to develop a multisectoral, patient-centered primary care council. Since July 2014, this council, consisting of English-speaking, Spanish-speaking, and Mien-speaking patients, as well as clinic staff and medical providers, has met on a monthly basis to explore patient-identified challenges within the clinic environment and to develop pilot projects in partnership with the clinic to address those challenges. The standardized Clinician and Group Consumer Assessment of Healthcare Providers and Systems scores for our clinic have improved by approximately 12% per year over the 3 years of the council's existence. The council has created a unique pathway through which the clinic can continue to engage with patient perspectives for enhancing the primary care experience. Over the past year, the council has been asked to comment on the development of quality improvement processes that have been mandated in our clinic through our safety net Medicaid waiver program. This includes the introduction of universal screening for depression and substance use, addressing our patients' sexual orientation and gender identification, and developing the messages to protect and support our undocumented patients. The council feedback has been reported to our Board of Trustees and the health system administration, who have used many of their suggestions in developing these programs.

The research approach used in this study demonstrates the strength of engaging with diverse perspectives in developing,

executing, and analyzing the results of a qualitative study. The study's findings highlight the importance of hearing the patient perspective as a component of developing a PCMH. In particular, findings highlight the added impact of linguistic differences between patients and clinic providers and staff. Understanding the challenges experienced by linguistically and culturally diverse patient communities has important implications for medical practice and education. While the themes highlighted by patient participants have been touched on in the literature, additional qualitative and quantitative research is needed to develop pragmatic methods to address key issues such as provider-patient interactions, the differential experiences of non-English-speaking patients, and the added effects of residency training programs on patients. Translating such research into practice is equally vital. Already, curricula that incorporate recognition of language and cultural differences into residency training are described in the literature [29,30]. In addition, the preference for language concordance with providers among patients who speak a language other than English suggests the importance of supporting medical training for physicians and staff who speak languages other than English and of developing methods to support such providers to practice within linguistically diverse settings in the United States [31].

Shim highlights that low-resource health care settings are simultaneously "more likely...to serve patients who lack significant cultural skills" to navigate the health care setting and more likely to be subject to the constraints of resources and time that would allow providers to "help patients become better participants in their own care" [13]. This situation is certainly true of LHAMC, where the hectic, packed schedules of attending physicians and residents necessitate 20-minute patient visits; such constrained visits are made even briefer when an interpreter is involved, cutting the amount of actual communication in half. The potential benefits of intentionally supporting effective patient engagement with the health care system and the individual doctor-patient interaction—in terms of saving time, money, and health in the future—suggest the need for safety net settings such as LHAMC to lengthen the patient visit time, allowing for provider-patient relationships to deepen even when a third party is necessary to broker a language gap. Incorporating the patient perspective—beyond isolated results from overarching patient satisfaction survey data—into every aspect of the clinician's role can enhance the patient's ability to fully engage with a primary care visit, giving the patient his or her best chance to benefit from that experience.

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Conflicts of Interest

None declared.

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Abbreviations

CHC: cultural health capital

LHAMC: Lakeview Hospital Adult Medicine Clinic

PCMH: patient-centered medical home

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