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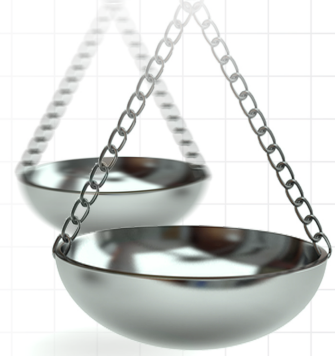

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Implementing a pragmatic framework for authentic patient–researcher partnerships in clinical research

In response to the creation of the Patient-Centered Outcomes Research Institute in 2010, researchers have begun to incorporate patient and family stakeholders into the research process as equal partners, bringing their unique perspectives and experiences to the table. Nonetheless, there is a dearth of literature around how best to engage patients and families and many barriers to doing so effectively. This paper outlines a pragmatic framework of collaborative engagement and partnership between research investigators and patient and family advisors from existing patient and family advisory councils (PFACs) at an academic medical center. This framework includes the role for each party throughout the clinical research process (launch, hypothesis, specific aims, measures/methods, results, interpretations/recommendation and dissemination).

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The decision to include patients as active participants in scientific research is a relatively new concept. The role of patients in research has traditionally been as study subjects, not peer collaborators. This method fits with the long-established paternalistic model of medicine, but has not yielded the answers clinicians and patients are looking for to make informed decisions about providing and receiving patient- and family-centered healthcare [1–3].

The Patient Protection and Affordable Care Act of 2010 mandated the creation of the Patient-Centered Outcomes Research Institute (PCORI) in order to advance comparative effectiveness research (CER) targeted to the needs of consumers and providers of healthcare. Specifically, PCORI promotes the assessment of prevention, diagnosis and treatment options; improvement of healthcare systems; communication and dissemination of research; addressing of health disparities and acceleration of patient-centered outcomes research (PCOR) and methodological research [4]. The backing for this new institute comes from the promise of fostering research processes and outcomes that are more relevant to the needs of patients, a core concept of patient- and family-centered care. To achieve these aims, PCORI supports research that incorporates patient and clinical stakeholders into the research process, from beginning to end [1–2,4–5].

Including patients or family caregivers as active stakeholders in research broadens the scope of scientific inquiry by bringing their unique experiences of care to the table. Among many advantages, patient-centered research enables the selection of appropriate and efficient methods, interventions that are tailored to patient needs and the spread of knowledge using patient-friendly dissemination channels. The role of patients and their caregivers in clinical research must shift from an approach that utilizes them solely as research objects to a process that engages them as equal partners in research.

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There are a growing number of studies that include various levels of patient or consumer involvement and confirm the benefit of a broader patient role [6–10]. Models have been proposed that begin to outline methods to engage numerous relevant stakeholders in patient- and family-centered research [7–9,11–18]. These frameworks describe guidelines for engaging research stakeholders, including patients, clinicians, policymakers, industry members and payers/purchasers. Many of these models outline potential engagement of various stakeholders, without outlining clear roles and best practices for executing engagement at each stage [11–13]. While useful, these models recommend blanket methods for engaging all stakeholders and, in doing so, fail to address the specific challenges of engaging patients.

The literature that focuses on patient engagement lacks concrete and comprehensive guidance for engaging patients as research partners [6]. Best practices of participatory research emphasize the importance of relationship building but offer little guidance beyond how to establish this initial partnership [8,15]. Other literature focuses solely on including the patient perspective via patient-reported outcome measures collected in surveys [16]. A systematic review of patient-engagement studies found that the majority limited patient engagement to focus groups, interviews or surveys, rather than inclusion in the research team. This review of 142 studies concluded that engagement is feasible in most settings but methods are needed to minimize tokenism and address logistical challenges [6].

PCORI outlines a conceptual model of PCOR, including a framework of foundational elements, supporting PCOR principles and outcomes for successful patient-centered research [14]. While the PCORI conceptual model includes actions to be taken to move from foundational elements to desired outcomes, these actions remain on a high level. This and other patient-engagement models do not elucidate how researchers who have never partnered with patients during the research process can execute patient engagement throughout the life of a study [12–14,17].

To facilitate successful authentic patient engagement, research institutions must harness existing structures that effectively engage patients and their families. Hospitals around the country are embracing patient- and family-centered care by creating and utilizing patient and family advisory councils (PFACs) [19]. PFACs are comprised of patients and family members that have received or observed care at the hospital and are then screened, interviewed, and oriented to their role as patient and family advisors (called PFAs or advisors). PFAs participate in operational decision-making as members of hospital committees, are engaged in quality improvement projects and are often involved in training staff on PFCC principles. More recently, PFAs have begun being sought to be peers within research teams.

Engaging PFAs is advantageous because they effectively communicate their perspectives and are oriented to the hospital, trained to represent the broader experience of patients/caregivers similar to themselves, and, if done correctly, represent a variety of patient and family backgrounds and experiences. PFACs are integrated into the structure of the organization and have operational leadership who support them, including providing access to institutional resources such as confidentiality and protection of research subjects training. Nass *et al.* summarize barriers that frequently prevent the engagement of patients as active partners, including power imbalances between researchers and patients/families, lack of organizational resources, difficulty in recruiting relevant patients, self-selected patients who are not representative of diverse patient populations and a lack of patient familiarity with scientific and medical terminology [18]. We believe that PFA involvement in research directly addresses many of these barriers, which typically preclude high levels of patient engagement in research.

The purpose of this paper is to describe a framework of collaborative engagement between investigators and PFAs and to identify determinants that enable this research approach at academic medical centers (AMCs). This pragmatic framework outlines the steps to partnering with an established PFAC throughout the clinical research process; for this paper, it has been specifically tailored to CER but is designed to be adaptable to and has been implemented with other approaches to clinical research. We describe operational roles and actions for the involved parties through each of the main stages of a research study, providing an illustrative example from our own work.

Our framework builds on the existing literature on patient engagement by providing practical steps to engage patients as equal members of the research team. First, it focuses specifically on patient/caregiver stakeholders and the unique guidelines needed to engage these stakeholders. Second, our framework seeks to translate broad theories and principles behind successful patient engagement into practice by outlining steps that guide researcher and patient actions through each stage of research. This approach is built on the same principles of trust, honesty, colearning, transparency, reciprocal relationships, partnership and respect outlined by PCORI [14]. Our framework seeks to operationalize these attributes, providing concrete steps that embody these PCOR principles. Finally, our novel approach has applicability to the growing number of institutions that have well-established PFAC programs, making it an important addition to the literature.

Pragmatic framework for authentic patient–researcher partnerships in clinical research

By clearly outlining the roles of a patient–researcher partnerships, we are laying out a framework for this new approach to research (see Table 1), which is designed to be put into practice by AMCs with active PFACs. We have outlined each stage of the research process and the actions taken by researchers and PFAs, as well as the facilitating PFAC leader (in Box 1), to ensure equal and collaborative participation from both parties.

This framework is built on the Brigham and Women's Hospital (BWH) Center for Patients and Families' experience engaging PFAs in research. The outlined processes have been implemented in multiple clinical research studies, including those involving a wide range of clinical fields (e.g., sleep apnea, care transitions, geriatric fall prevention) and in inpatient and outpatient settings. This framework has supported patient/family engagement in a wide range of study designs, such as stepped-wedge and randomized control trials. BWH studies designed to engage PFAs using this framework have received funding from multiple organizations promoting patient/family engagement in research, including PCORI and the Agency for Healthcare Research and Quality. This framework has been developed and adjusted based on lessons learned from our experience engaging advisors in research.

To help illustrate our framework, we will provide examples at each stage of implementation of a PCORI-funded study, "Relative patient benefits of a hospital Patient-Centered Medical Home collaboration with an Accountable Care Organization (ACO) to improve care transitions" (Easygrants ID 811; called the 'Care Transitions' study henceforth), led by Dr J Schnipper. The Care Transitions study's long-term goal was to assess the benefits and harms of a novel collaborative transitions intervention between the Partners Healthcare Pioneer ACO and several of its patient-centered medical homes. To do so, the main aims of the study were to: develop, implement and refine a multifaceted, multidisciplinary transitions intervention; evaluate the effects of this intervention on post-discharge adverse events, functional status, patient satisfaction and emergency department and hospital utilization within 30 days of discharge; and understand the barriers to and facilitators of successful implementation of this intervention across practices. Staff from the Center for Patients and Families and an experienced PFA were engaged during the proposal synthesis stage to develop a patient-engagement plan. Additional PFAs were engaged from the launch to the study's current stage of results analysis as of February 2016.

In order to provide the necessary infrastructure to recruit PFAs whose medical problems and lived experiences of care are congruent with the research being conducted, the AMC's research leadership must form a partnership with existing PFACs and their PFAs. To do so, researchers need to seek out the leader operationally responsible for the PFACs; at BWH, this is the executive director of the Center for Patients and Families, whose operational role includes leading PFACs, recruiting advisors and supporting their engagement in hospital activities. A natural alliance can be established between research and PFAC leadership to recruit and train interested PFAs.

To begin this process, the principal investigator (PI) summarizes the proposed research study to the PFAC leader and describes ideal attributes of PFAs. The PFAs can be advisors that work within a certain clinical field that are brought in once the researcher has requested them, or they can be a group that is already working with the researcher; regardless, PFAs will participate only as part of the research team and cannot be research subjects once engaged in the study. The ideal situation is to involve PFAs as early as possible in the process, even before a proposal has been written and submitted or the research question decided upon, but advisors can still be matched later in the research process if needed. The PFAC leader creates or uses a narrative matrix of the PFA's lived healthcare experiences. This matrix is a detailed, qualitative database designed to keep track of all interested PFAs and their stories, thus facilitating the pairing of PFAs with relevant clinical research projects. This matrix can also record logistically helpful information such as past participation in research studies and which advisors have received annual Institutional Review Board trainings.

The PFAC leader acts as a neutral facilitator supporting the implementation of the framework. The facilitator role is especially important initially as the researchers learn how to engage the PFAs in each phase of the research study (see Box 1 for details about the PFAC leader's role throughout the study). The PFAC leader coordinates the initial team meeting and ensures appropriate accommodations are made to facilitate PFA involvement, such as a frequency of and location for meetings which can be sustained, flexibility to address specific patient/family needs and methods of communication which work for PFAs (which may not be traditional researcher means of communication). The PFAC leader helps researchers understand that involving PFAs successfully in research requires acknowledging and addressing their needs, including assessing their level of experience with research, ensuring their understanding at each stage and providing an adequate stipend to support their involvement [10,,20–21].

Table 1. Pragmatic framework for authentic patient–researcher partnership in clinical research.

Research phases	Researcher role	Patient/family advisor role	Collaboration/dialogue together

Launch	– Establish shared decision-making
	– Provide tutorial on:
	  – Rigor
	  – Validity
	  – Reliability
		– Participate in partnership building discussion
		– Learn research principles
			– Broker partnership
			– Discuss research principles and proposal

Hypothesis	– Explain why the problem is significant and why you study this field
	– Ask advisors: does this resonate with you?
		– Provide feedback:
		  – Is this research question important to us?
		  – Will its answer benefit patients in realistic situations?
			– Determine:
			  – What is the right research question?
			  – What outcomes are important to advisors and researchers?

Specific aims	– Share current research landscape for the study question
	– Ensure realistic aims are set
		– Provide feedback:
		  – Will these aims resonate with patients' lived experiences of research area?
		  – Will aims produce useful outcomes for patients?
			– Discuss:
			  – Why are these aims important (to research field and patients)?

Methods/measures	– Review how to measure outcomes
	– Provide tutorial on:
	  – Bias
	  – Confounding
	  – Chance
	  – Tools/study attributes
		– Learn about study methodology
		– Ensure study intervention and methods are sensitive to needs of patient population
			– Explore methods that produce valid research
			– Discuss:
			  – How will intervention and methods affect patients?

Results	– Present data collected and analysis
	– Populate tables with actual data
	– Provide tutorial on:
	  – Mock up practice tables to review understanding data
	  – Present and explain results
	  – Translate numbers/values
		– Continue engagement in study by preparing to understand results
		– Provide feedback to refine methods/intervention as needed
		– Apply actual data to learned research framework
			– Assess validity of results

Interpretations/ recommendations	– Interpret scientific language
	– Provide tutorial on:
	  – Threats to external validity
	  – Evaluation of applicability/ generalizability
		– Translate/interpret data and its application into laymen's terms	– Draw scientific conclusions and interpret in laymen's terms

Dissemination	– Discuss: what have we learned? – Publish scientific paper	– Use experiences to validate message – Publish results to targeted popular and social media	– Message for and share results with clinical, patient and community stakeholders

Box 1. Patient and family advisory council leadership role in facilitating patient–researcher partnerships.

In the initial stages of this partnership and throughout the life cycle of the research study, the patient and family advisory council leader plays an important role in facilitating and supporting the application of the Pragmatic Framework

Role in preparing for launch

- Meet with PI to determine scope and needs of research study
- Identify PFAs with lived experiences that match area of research
- Determine PFA interest in participating in research study
- Ensure appropriate accommodations have been made to facilitate a partnership (meeting times, communication methods, compensation for PFAs, among others)
- Coordinate and facilitate initial meeting between PFAs, PI and other researchers

Role in launch

- Promote relationship-building between PFAs and researchers by leading conversations about why each is engaged in this research
- Support PI, researchers and PFAs to develop plan for PFA involvement in the research study
- Ensure expectations for each role are clearly communicated and agreed upon
- Prepare researchers prior to meetings by providing tutorial examples, sharing lessons learned and answering questions about PFA engagement

Facilitating role throughout (as co-investigator or consultant)

- As needed, attend all or periodic meetings to participate:
  - Ask questions to promote sharing and clarification of information the PFAs may not know or understand
  - Support researchers to check that PFAs understand their role and research content at each stage
- Check in regularly with PFAs and researchers individually to ensure:
  - Researcher needs are being met by PFA engagement
  - PFA feedback is being integrated in a meaningful way
- Support troubleshooting and identifying solutions to partnership challenges

PFA: Patient and family advisor; PI: Principal investigator.

Prior to the launch of the Care Transitions study, the PFAC leader provided guidance to the PI regarding practices that would best support PFA involvement. This included recruiting an experienced advisor to be engaged early on and working together to provide input into the patient-engagement plan in the grant proposal. The PFAC leader recommended advisors be paid US$50/h and offered reimbursement for expenses incurred to attend meetings. The patient-engagement plan included monthly meetings between the PI, research staff and PFAs as well as PFA representation on the study Steering Committee. The PFAC leader then led the recruitment of six advisors based on relevant experiences of care as a patient or caregiver (see Table 2 for PFA characteristics) and met with the researcher prior to meeting as a group, in order to brainstorm aims and expectations for each stage of the study.

Launching the research study

During this initial phase of the research study, the newly formed team must build a foundation for mutual understanding, collaborative communication and shared decision-making throughout the study. To broker this partnership, it is vital for the researchers, PFAC leader and PFAs to clearly discuss the research process and to establish a common level of language and wording, in order for the PFAs to understand more clearly and be comfortable participating. From the beginning, researchers should actively listen to and value the PFAs' input in order to facilitate the formation of this partnership. In the Care Transitions study, one way researchers showed this was by eliminating degrees from nametags and introductions, thereby signaling that all contributions were equally valid.

In this stage, components of a grant application are introduced and basic concepts and tenets of clinical research including rigor, validity and reliability are explored. A basic understanding of the scientific process is necessary for PFAs to participate in research. Varying degrees of knowledge and experience with research concepts will require researchers to adopt a new role as teachers to effectively convey these and other concepts, such as power, bias and chance, many of which can be addressed as the research progresses (see Supplementary Data for an example of the tutorial process). The PFAC leader should be available prior to the launch to discuss tutorials with researchers; the PFAC leader should also guide the researchers to clarify concepts and provide further examples during the meetings.

The PFAC leader can further facilitate initial conversations and trust building by asking questions that an advisor might not know the answer to. In many

Table 2. Patient/family advisor characteristics from Care Transitions study.

Characteristics	Patient/family advisors (n)
Role:	
– Patient	3
– Family/caregiver	3
Sex:	
– Female	4
– Male	2
Age (years):	
– 40–50	1
– 51–60	1
– 61–70	0
– 71–80	3
– 81–90	1
Ethnicity:	
– Hispanic	0
– Not Hispanic	6
Race:	
– African–American/black	2
– Caucasian/white	4
– Other	0
Education:	
– Less than high school diploma	0
– High school graduate	1
– Some college	1
– College graduate	2
– Higher degree	2

Patient and family advisors serving on the Care Transitions study (funded by the Patient-Centered Outcomes Research Institute) represented broad experiences of healthcare and patient and family advisory council (PFAC) membership. Because this study encompassed inpatient and outpatient care as part of the Partners Accountable Care Organization, advisors were recruited from PFACs at two hospitals and an outpatient clinic within this Accountable Care Organization. Patient and family advisors had experience as part of:
– Brigham and Women's Hospital (BWH) emergency department PFAC;
– BWH Enterprise PFAC;
– BWH Shapiro cardiovascular/renal transplant PFAC;
– BWH South Huntington Medical Home PFAC;
– Massachusetts General Hospital (MGH; MA, USA) Ambulatory Practice of the Future Care Alliance;
– MGH Cancer Center PFAC;
– MGH General Hospital PFAC.

cases, PFAs may not understand why researchers would seek their opinion or what they can contribute with their potentially limited knowledge of research. During the Care Transitions study, it helped PFAs to learn what inspired the researchers to work in this field. The PFAC leader can facilitate this by asking: Why is the researcher conducting research about this topic specifically? These conversations provide the motivational drive for all members to partner together, as the researcher's answer is often closely connected to the realities of care that the PFAs may have experienced. This led the Care Transitions team to start each meeting by having one team member share their story and role, so that both researchers and PFAs had the opportunity to learn about one another.

Another important question the PFAC leader can ask the PI is: once the study is over and the results are disseminated, what happens when your peers read the article? Answers which describe the process of peer review and challenges to the conclusions of newly published research are often met with surprise from PFAs if they do not have a research background. As lay people, PFAs may come into this process regarding researchers as unquestionable experts; sharing this vulnerability helps even out the power dynamic, creates an allegiance between PFA and researchers and helps PFAs understand the importance of scientific rigor. After this conversation, the advisors in the Care Transitions study felt motivated to understand study fundamentals, as they grasped the importance of helping the PI create a rigorous study. Introducing the idea of using valid and reliable tools to do so will help make this more concrete for PFAs. To ensure understanding, it is always essential to explain each concept, why it is important to the research process and what it looks like to have a rigorous, valid and reliable study in practice.

To facilitate PFA involvement in the development of the research question for a CER study, the researcher provides exemplars to illustrate this approach (which can be adapted to other methodologies). PFAs should understand that CER studies seek to compare different treatments and their outcomes, with the goal of providing patients and clinicians with results that can help guide treatment decisions [22]. In the Care Transitions study, several examples were provided, such as a prostate study evaluating difference in outcomes of prostatectomy as compared with other options like watchful waiting [23].

It is also critical that PFAs understand how the framing of the research question can affect knowledge gained from the study. Discussions could include concepts like absolute versus relative benefit. Giving the example of oncology treatment that prolongs survival by 2 weeks without taking quality of life into account will help PFAs understand this difference. Understanding this concept will support PFAs to express what knowledge would be worth gaining based on their clinical experience. Having this conversation allows researchers to learn what PFAs want to know and gives PFAs meaningful input into the direction the study will take, while underscoring the importance of their feedback.

While the steps described in this section ideally occur simultaneously with the launch of the research

study and prior to the synthesis of the study's research question, relationship brokering and research tutorials are essential first steps to PFA engagement regardless of when PFAs join the research team.

Hypothesis

After the PFAs have been oriented to general research concepts, it is the researcher's undertaking to teach the PFAs about the specific area of interest. Together, the team determines the most appropriate and useful research question to be studied and hypothesis to be tested. The question must resonate as a significant area of study for both the researchers and the PFAs, and represent the interests of the patient population. To do so, the researchers must get the PFAs' input on questions that will direct the focus and mechanics of the study. The team works together to ensure that the research question yields the most beneficial answer for the constituency the PFAs represent and the primary outcomes will address realistic situations.

While we believe engaging advisors during the design of the research question is one of the most significant and novel ways the patient/caregiver voice can be included in research, we also recognize that this can present logistical challenges, as grant request for applications may require specific criteria or researchers may anticipate their next project. With the support of the PFAC leader, researchers can actively manage this tension by explicitly explaining to PFAs which components are negotiable and which are predetermined. Researchers should make an effort to remain open to and seek input on fundamental aspects of the study, as PFAs often offer insights that are not anticipated or expected.

The PFAC leader and original advisor initially provided input on topic solicitation and hypothesis framing during the Care Transitions proposal writing stage. They gave feedback on the importance of this topic and whether the proposed research question was important to the relevant patient population. Once the proposal was accepted and the other PFAs recruited, the hypothesis was revisited, explained and placed in context of the existing work on care transitions.

Specific aims

After the research question is agreed upon, the researcher should fine tune the hypothesis based on a literature review. It is important for the researcher to determine what information is already available and where the study should aim to add to the field. It is also the researcher's responsibility to collect PFA perspectives on specific aims which will elicit the information they are interested in. As such, the researcher ensures that the study is focused on both the interests of the affected patients and that the right aims are outlined to test the hypothesis.

For the Care Transitions study, this stage took the form of sharing existing discharge practices within the Partners ACO so that PFAs could understand the baseline practice and outcomes; this helped PFAs understand why the general aims had been chosen by the researchers. Though the general aims of the study were predetermined, they remained broad at the outset so advisor input focused on narrowing their scope by revising specific aims to include those that were specifically meaningful in the lives of patients. For example, the aims of reducing clinically meaningful measures such as post-discharge adverse events and rehospitalization rates were supplemented with the aims of improving the ability of the patient to return to work, if applicable, and decreasing the extent to which patients reported feeling like a burden to family and other caregivers.

Methods/measures

During this stage, the study methods and outcome measures are explored together by the researchers and PFAs. Researchers should begin by teaching PFAs about additional research concepts, which will prepare the PFAs to discuss patient-facing interventions and data collection methods, such as surveys and interviews. The key is for PFAs to be inspired by how challenging it can be to produce high-quality research and to have a central role in molding the way researchers interact with patients when implementing study methods.

The researcher begins by teaching the PFAs about chance, bias and confounding (see Supplementary Data for a tutorial example), the major threats to the internal validity of a study. It is important for PFAs to understand that researchers must be aware of and address each of these factors when planning a study's methods and measures. Defining each concept, giving concrete examples of how each occur in research and describing how to lessen the impact of each of these factors during the study design and analysis ensures that the PFAs grasp these concepts. Explaining these concepts in plain language builds a common understanding between PFAs and researchers. In the Care Transitions study, some PFAs felt they did not need to know this information in order to fulfill their role; however, the PFAC leader felt that a full understanding would ensure PFA confidence in their grasp of the study, allow them to provide informed input regarding the analysis plan and prepare them for the results and interpretations stages.

When the PFAs are comfortable with these concepts, researchers and PFAs collaborate to determine methods and measures that will produce a high quality study and accommodate patient needs. Researchers lead this pro-

cess, proposing methods and measures and are responsible for making sure each PFA understands and is able to contribute their perspective as to how these choices will impact patients. This process should include the design or refinement of the study intervention itself. Even a predetermined clinical protocol can have room for the PFA perspective; for example, the PFA might weigh in on how communication with the patient is approached. In other cases, the PFA can play an active role in the development and refinement of the intervention itself, providing feedback on the needs of the patient population and pilot testing versions throughout the process. PFAs should also be given the chance to pilot patient-facing data collection instruments for ease of use, clarity in instructions and language and applicability to the patient population. Together, the researchers and PFAs refine the study intervention, methods and measures to allow for the best possible interactions with patients, while maintaining the integrity of the study.

Advisors played an important role in refining the Care Transitions intervention, participating in decision-making, document and tool tailoring and training of research staff executing study enrollment. PFAs gave input on specific components of the interventions, such as 'Who would you want to be told to call with questions/concerns after discharge and what is the best way to present this information in the discharge summary?' They were provided with all patient-facing documents and asked to review and provide feedback on clarity and presentation. PFAs also refined tools created for clinicians involved in the intervention, such as a Discharge Advocate Checklist, which was designed to remind clinicians to complete and document important tasks at discharge. PFAs supported research implementation, as well. The Research Assistant who enrolled patients in the study performed a mock intake interview with a PFA in front of the group; the PFAs then discussed and provided feedback to the Research Assistant on patient-friendly language and behaviors.

PFA input also proved to be essential to the Care Transitions' data collection methods. PFAs played an active role in refining the proposed survey content and administration process. Advisors pilot tested the questions and ensured that they were clear and direct, providing feedback that led to meaningful edits. PFAs felt that it was important to assess which piece of the intervention patients liked most, rather than solely assessing the most effective component, as proposed by researchers. They believed this measure would reveal intervention components that patients found most valuable, least intrusive, and easiest to adhere to, among other possibilities. This new perspective led the team to add a focus group with intervention patients as a qualitative measure to assess which intervention component patients liked most. The focus group with six randomly selected patients who participated in the intervention was led by a PFA and the PFAC leader, with a qualitative researcher listening in and study staff transcribing the dialogue for further qualitative analysis.

Results

To ensure PFAs continue to provide feedback and are prepared to understand the results of the study, regular meetings should be planned throughout the implementation period of the study. These meetings allow researchers to continue to solicit the patient/family perspective on barriers that may arise during implementation and support continuing education for the advisors. Researchers can facilitate PFA engagement and understanding by creating tables and graphs with mock results or current data and have PFAs practice reading and summarizing data from these tables. It is also important to explain the meaning of each component of these tables and concepts that arise in interpreting them, such as n collected, CIs, adjusted analysis, p-values, correlation and causation. These meetings also serve to keep advisors updated on progress, allowing them to build realistic expectations for the study results.

The Care Transitions study is currently in this stage of the study, as enrollment has finished and results are being analyzed. Meetings during the intervention period covered enrollment progress, troubleshooting of intervention challenges and monitoring input from patient participants. Selected patient subjects were interviewed by the team as a means to gather qualitative data and feedback on the intervention; concerns or issues that surfaced in these interviews were addressed by researchers and PFAs together. PFAs also practiced reading preliminary results and discussing their interpretations in preparation for the final stages of the study.

Once the data have been collected and analyzed, the research team prepares tables and graphs of the findings. The researchers present the data to PFAs in an organized way and explain the meaning of each result in order to support the PFAs' understanding. To do so, link values shown in the tables and graphs back to the concepts PFAs were taught during the methods and measures phase. The PFAs now seek to apply the hard data to the research framework they have learned throughout this process, considering whether the findings answer the research question.

Interpretations/recommendations

After the findings have been organized, the research team interprets the results for applicability and generalizability, determining recommendations for clinical practice or further research. The researchers explain their interpretations to the PFAs, describing factors that

threaten the study's external validity and explaining the applicability and generalizability to various populations or contexts. Together, researchers and PFAs should discuss limitations and opportunities for future research.

The PFAs' understanding of this is important, as it will help them to translate the conclusions and recommendations into lay terms to allow for the broader dissemination of the study findings, especially to the populations of patients affected by the results. The researchers and PFAs work together to create a narrative that the PFAs can use to explain the study design, methods and results in an accessible way.

While the Care Transitions study has not yet reached this phase, the patient-engagement plan outlines a process for translating results into a rigorous and accessible message for dissemination. Researchers will present the results and their scientific interpretations and give PFAs a chance to provide input. The team will discuss whether the conclusions of the study make sense or are as expected. The PFAs will provide input on how to best explain the results to other patients and the public so that they resonate and are comprehensible. Together, the PFAs and researchers will decide how to convey the study's conclusions for both scientific and popular audiences.

Dissemination

Once the results have been interpreted, the researchers and PFAs work together to create a dissemination plan that includes professional and lay publication. Researchers will present their plan for writing manuscripts for peer-reviewed journals and presenting with PFAs at professional conferences. In addition to the usual scientific dissemination channels, our framework provides a unique avenue for the results to be disseminated among lay people. PFAs will recommend media outlets that are accessible to those outside the research community, such as targeted popular media, news outlets, state and national stakeholder associations and patient affinity organizations. In these publications, study findings will be shared in a narrative form, using language determined by the PFAs and researchers. As results are disseminated, researchers and PFAs should include and give credit to the others' contributions. Through these complementary dissemination methods, the study conclusions will reach scientists, researchers, medical professionals and a targeted portion of the general population who may be most interested in or affected by the results.

While final decisions cannot be made until this phase of the study, conversations about dissemination can begin earlier in the research process. This ensures that researchers have considered how they will present methods or results and that advisors have actively thought about the best means to communicate results to the public prior to the end of the study. The Care Transitions study has begun preparing for this stage by discussing potential avenues for dissemination and what information the team hopes to convey in scientific and lay publications.

Discussion

As detailed earlier, this framework is forged from our experiences engaging PFAs in research through the BWH Center for Patients and Families. This model of engagement has been implemented successfully in a variety of settings, study designs and clinical specialties. In its ideal form, PFA involvement begins prior to the study launch, as described by our framework, but PFAs have also been meaningfully integrated later in the process. At present, the BWH studies using this framework are still in progress. The Care Transitions study is furthest along and in the results analysis phase; as such, formal evaluation to measure the impact of PFA engagement in the research process has not been conducted, which we acknowledge as a limitation to our framework. Nonetheless, we are encouraged by the framework's success in practice and continued requests by PIs for PFA involvement in their projects.

Furthermore, others have proposed evaluative criteria to determine the success of stakeholder engagement in the absence of quantitative analyses. The PCORI conceptual model of PCOR outlines a culture of patient-centeredness and meaningful and effective partnerships between team members as the near-term outcomes of successful stakeholder engagement [14]. Lavallee *et al.* identified respect, trust, legitimacy, fairness, competence and accountability as measures for optimal stakeholder engagement [24]. In our experience, these factors are critical in determining the success or failure of PFA engagement in research. To assess whether our framework has been successful by these measures, we have collected qualitative feedback from PFAs and researchers that illustrates their perspectives on the culture and values fostered by this process. For example, in the Care Transitions study, advisors reported positively on key measures of partnership between them and the researchers: they felt the expectations outlined were met, communication was respectful and open, their input was valued throughout the study and changes were made based on their feedback. Similarly, researcher feedback also reflected a positive experience. They felt PFAs contributed meaningful insights that resulted in positive changes to every stage of the study, were actively engaged and were important members of the research team.

We also recognize the favorable circumstances at Brigham and Women's Hospital, which allowed us to build a robust PFAC program with a diverse and talented group of PFAs, thus facilitating this type of engagement.

BWH's institutional prioritization of patient- and family-centered care and our location in Massachusetts, which is currently the only state that legislates a requirement for hospitals to create PFACs, encouraged the early adoption and growth of this feature. While Massachusetts does not mandate PFAC involvement in research, the law has promoted the growth of PFACs into robust and active participants in hospital activities, which reinforces PFAC ability to engage in activities like research. Furthermore, due to the visibility and success of the BWH PFAC program, researchers have sought out the involvement of PFAC members, contributing to our ability to integrate PFAs successfully into research projects.

Nonetheless, because PFACs have become increasingly widespread at AMCs, we see wide applicability of our framework. Legislation requiring PFACs is not a requisite to their growth, as evidenced by the more than 2000 hospitals that have launched PFACs since 1990. As PFACs become well established at hospitals around the country, they increasingly participate in research [25,26]. With the creation of PCORI, incentives to create and use PFACs to engage PFAs and patient and families in research have increased; our framework supports institutions to do so in a comprehensive and successful way. We also see opportunities for further study of our framework, including a formal analysis of its impact on the research process.

Conclusion

With passage of the Affordable Care Act, there has been a significant increase in institutional and financial support for patient/family engagement in research. We have outlined a pragmatic framework to enable successful and impactful collaboration between patients and researchers. As the patient/family perspective is given equal footing to other stakeholder perspectives and their lived experience is incorporated throughout the research process, research outcomes will better meet the needs of relevant patient populations and increase patient-centered practices in research and care delivery. Going forward, we see additional opportunities to expand current healthcare institutional and research engagement of patients and families as well as the need for further studies on the efficacy of engaging them in different ways.

Future perspective

Despite the significant growth in the number of PFACs and their meaningful involvement in healthcare organizations around the country over the last 10 years, PFA engagement in research remains limited [2,25–26]. As PFAC programs continue to grow and mature, this improved infrastructure will support increased PFA involvement in research. PCORI's leadership in funding and promoting patient-centered research will continue to encourage CER and other patient-centered outcomes research that includes patient and family stakeholders as part of the research team. PCORI's recent development of various networks to support sharing of research methodology, data and lessons learned, such as the National Patient-Centered Clinical Research Network and the Evidence to Action Networks, and their dissemination of results from early PCORI-funded studies will further increase the efficacy and profile of this approach[27,28]. Concrete steps for engagement, as outlined in our pragmatic framework, will provide researchers with a blueprint to effectively engage patients and families in research.

Consequently, we hope the next 10 years will see a shift toward patient- and family-centered CER, which, through the exploration of research questions directly relevant to patient and family lived experience, will lead to findings with the potential to directly impact care in novel and dramatic ways. The dissemination of these results in accessible consumer-oriented media will empower patients and families to educate themselves and engage in their care to a new extent. The goal and challenge will be to translate this research and its results into the delivery of patient- and family-centered care on a systemic level. It will be the responsibility of healthcare leaders to push for the integration of new standards and practices into care while keeping patient and family values and needs at its center.

Supplementary Data

To view the supplementary data that accompany this paper please visit the journal website at: www.futuremedicine.com/doi/full/10.2217/pme-2015-0023

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Executive summary

- While there is growing recognition that patient/family engagement in research is critical to producing results which best meet clinical needs, there is limited literature on how to do so effectively.
- There are numerous barriers to engaging patients/families effectively in research.
- We propose a framework to engage patient and family advisors (PFAs) from established patient and family advisory councils (PFACs) in research, an approach that addresses many of these barriers.

Pragmatic framework for authentic patient–researcher partnerships in clinical research

- Researchers work with PFAC operational leadership to identify PFAs that are a good fit for research study.
- The PFAC leader facilitates initial logistics and development of partnership between researchers and PFAs.

Launching the research study

- Researchers and PFAs set standards for communication and understanding.
- Researchers introduce general research concepts to PFAs to prepare them to understand the research process.

Hypothesis

- Researchers ensure PFAs have a broad understanding of research topic.
- Researchers and PFAs work together to develop research question that meets needs of patient constituency.

Specific aims

- Researchers and PFAs refine hypothesis to target aims that add to existing research and meet patient needs.

Methods/measures

- PFAs continue to learn about research concepts that ensure meaningful outcomes.
- Researchers and PFAs determine and refine methods/measures that accommodate patients and ensure valid results.

Results

- Regular meetings throughout the implementation period keep PFAs engaged in the study and informed of evolving results.
- Researchers organize and present results to PFAs, helping PFAs to apply previously learned concepts to understand the meaning and validity of the results.

Interpretations/recommendations

- Researchers explain generalizability and applicability of results.
- Researchers and PFAs develop scientific recommendations and translate these into narratives for lay people.

Dissemination

- Findings are shared via traditional research means and in relevant lay media identified by PFAs in narrative form.

Discussion

- This framework is built on our experience engaging Brigham and Women's Hospital PFACs in research but has broad applicability to other academic medical centers with PFACs.
- Limitations include a lack of formal analysis of the impact of the framework and favorable circumstances that led to its development and implementation.

Conclusion

- This framework provides a pragmatic outline to incorporate PFAs into research as equal partners.
- Further research is needed to examine the efficacy of various strategies for engagement.

References

Papers of special note have been highlighted as:
• of interest; •• of considerable interest

1 Frank L, Basch E, Selby JV. The PCORI perspective on patient-centered outcomes research. *JAMA* 312(15), 1513–1514 (2014).

•• **This perspective piece outlines the philosophy behind the Patient-Centered Outcomes Research Institutes (PCORI's) requirement for patient engagement in research, its added value and the work PCORI is doing to further explore and understand methods of patient engagement that uphold scientific rigor and increase effectiveness of research undertaken.**

2 Coelho T. A patient advocate's perspective on patient-centered comparative effectiveness research. *Health. Aff. (Millwood)* 29(10), 1885–1890 (2010).

• **Coelho shares his personal experience as a patient and long-time advocate, explaining the importance of patient-centered comparative effectiveness research that engages patient stakeholders and his belief that comparative effectiveness research will lead to significant improvements in patient care and education.**

3 Institute of Medicine. *Crossing the Quality Chasm: a New Health System for the 21st Century*. The National Academies Press, Washington, DC, USA (2001).

4 Selby JV, Beal AC, Frank L. The Patient-Centered Outcomes Research Institute (PCORI) national priorities for research and initial research agenda. *JAMA* 307(15), 1583–1584 (2012).

5 Selby JV. The Patient-Centered Outcomes Research Institute: a 2013 agenda for "research done differently". *Popul. Health Manag.* 16(2), 69–70 (2013).

6 Domecq JP, Prutsky G, Elraiyah T *et al.* Patient engagement in research: a systematic review. *BMC Health Serv. Res.* 14, 89 (2014).

•• **This systematic literature review found that patient engagement in research is feasible in practice but that gaps exist as to the best methods to ensure sustained, legitimate engagement throughout the process.**

7 Shalowitz MU, Isacco A, Barquin N *et al.* Community-based participatory research: a review of the literature with strategies for community engagement. *J. Dev. Behav. Pediatr.* 30(4), 350–361 (2009).

8 Bogart LM, Uyeda K. Community-based participatory research: partnering with communities for effective and sustainable behavioral health interventions. *Health Psychol.* 28(4), 391–393 (2009).

9 Devine EB, Alfonso-Cristancho R, Devlin A *et al.* A model for incorporating patient and stakeholder voices in a learning health care network: Washington State's Comparative Effectiveness Research Translation Network. *J. Clin. Epidemiol.* 66(8 Suppl.), S122–S129 (2013).

10 Forsythe LP, Ellis LE, Edmundson L *et al.* Patient and stakeholder engagement in the PCORI pilot projects: description and lessons learned. *J. Gen. Intern. Med.* 31(1), 13–21 (2016).

•• **Details lessons learned from early PCORI pilot studies, providing insight into the stakeholders engaged and how, stakeholder contributions and facilitators and challenges.**

11 Concannon TW, Meissner P, Grunbaum JA *et al.* A new taxonomy for stakeholder engagement in patient-centered outcomes research. *J. Gen. Intern. Med.* 27(8), 985–991 (2012).

• **Concannon *et al.* propose a taxonomy to identify stakeholders to engage in research, clarify roles and responsibilities at each stage of research and recommend steps for researchers to take to begin engaging stakeholders.**

12 Deverka PA, Lavallee DC, Desai PJ *et al.* Stakeholder participation in comparative effectiveness research: defining a framework for effective engagement. *J. Comp. Eff. Res.* 1(2), 181–194 (2012).

13 Hoffman A, Montgomery R, Aubry W, Tunis SR. How best to engage patients, doctors, and other stakeholders in designing comparative effectiveness studies. *Health Aff. (Millwood)* 29(10), 1834–1841 (2010).

14 Frank L, Forsythe L, Ellis L *et al.* Conceptual and practical foundations of patient engagement in research at the patient-centered outcomes research institute. *Qual. Life Res.* 24(5), 1033–1041 (2015).

15 de Wit MP, Elberse JE, Broerse JE, Abma TA. Do not forget the professional - the value of the FIRST model for guiding the structural involvement of patients in rheumatology research. *Health Expect.* 18(4), 489–503 (2015).

16 Wu AW, Snyder C, Clancy CM, Steinwachs DM. Adding the patient perspective to comparative effectiveness research. *Health Aff. (Millwood)* 29(10), 1863–1871 (2010).

17 Mullins CD, Abdulhalim AM, Lavallee DC. Continuous patient engagement in comparative effectiveness research. *JAMA* 307(15), 1587–1588 (2012).

18 Nass P, Levine S, Yancy C. Methods for involving patients in topic generation for patient-centered comparative effectiveness research: an international perspective. Research Priorities White Paper (PCORI-SOL-RPWG-001) for the Patient-Centered Outcomes Research Institute, Washington, DC, USA (2012). www.pcori.org/assets/Methods

•• **Nass *et al.* describe the importance of and process for engaging the public in research, proposing a patient-centered typology of engagement and outlining specific protocols for engaging the public at various levels.**

19 Herrin J, Harris KG, Kenward K, Hines S, Joshi MS, Frosch DL. Patient and family engagement: a survey of US hospital practices. *BMJ Qual. Saf.* 25, 182–189 (2016).

20 Patient-Centered Outcomes Research Institute. PCORI compensation framework for engaged research partners (2015). www.pcori.org/sites/default/files

• **PCORI details a framework for financial compensation for patients as engaged partners in research, providing the rationale for and guidance on fair compensation.**

21 Agency for Healthcare Research and Quality. Supporting patient and consumer involvement in patient-centered outcomes research. AHRQ Community Forum (2011). www.effectivehealthcare.ahrq.gov

22 Tunis SR, Benner J, McClellan M. Comparative effectiveness research: policy context, methods development and research infrastructure. *Stat. Med.* 29(19), 1963–1976 (2010).

23 Bill-Axelson A, Holmberg L, Ruutu M *et al.* Radical prostatectomy versus watchful waiting in early prostate cancer. *N. Engl. J. Med.* 352(19), 1977–1984 (2005).

24 Lavallee DC, Williams CJ, Tambor ES, Deverka PA. Stakeholder engagement in comparative effectiveness research: how will we measure success? *J. Comp. Eff. Res.* 1(5), 397–407 (2012).

• **Highlights the importance of and need for evaluative measures for stakeholder engagement and proposes six meta criteria for measuring its success.**

25 Lewis B. PFACs: where's the money? patient experience research report. The Beryl Institute, Bedford, TX, USA (2014). www.ipfcc.org/advance/topics

26 2015 PFAC Summary Report. Health Care for All, Boston, MA, USA (2015). www.ipfcc.org/advance/topics

27 PCORnet, The National Patient-Centered Clinical Research Network (2015). www.pcornet.org

28 Anise A. Our new evidence to action networks - helping awardees share knowledge. Patient-Centered Outcomes Research Institute, Washington DC, USA (2015). www.pcori.org/blog/our-new-evidence