Stakeholder Engagement Basics for Health Science Researchers: Putting the Pieces Together

March-June 2022, Virtual

Facilitator: Gay Thomas, GR Thomas Advisors LLC

Sponsored by CAIRIBU

Table of Contents

Series Overview 1
Participant learning objectives 1
Series facilitator information 1
Series agenda 2
Cohort information 3-4
Session 1 5
Registration form 6
Article: "Practical Guidance for Involving Stakeholders in Health Research" 7
Session 1 presentation 13
Session 2 20
Session 2 overview 21
Article: "A Case Study of Engaging Hard-to-Reach Participants in the Research Process: Community Advisors on Research Design and Strategies (CARDS)® 22
Article: "The Power of The Personal: Breaking Down Stereotypes and Building Human Connections" 31
Sample stakeholder engagement agenda 35
Session 3 37
Session 3 overview 38
Session 4 39
Session 4 overview 40
Article: "Patient-Engaged Research: Choosing the "Right" Patients to Avoid Pitfalls" 41

Collaborating for the Advancement of Interdisciplinary Research in Benign

Stakeholder Engagement Basics for Health Science Researchers: Putting the Pieces Together

Participant learning objectives:

- Identify stakeholder categories and groups most relevant to their program of research
- Understand how stakeholders can affect research and research outcomes
- Understand how stakeholder engagement could add value to your program of research
- Recognize important elements in a stakeholder engagement plan
- Describe key steps in planning stakeholder meetings
- Gain awareness of existing resources to support stakeholder engagement

---

Series Facilitator: Gay Thomas, GR Thomas Advisors LLC

- Gay is working with the CAIRIBU U24 Interactions Core to support and enhance our capacity to effectively engage stakeholders in benign genitourinary research.

- ***Why Gay?***
  - She is a stakeholder engagement consultant with over 30 years of experience in public policy, the private sector and health sciences research.
  - She has 20 years of experience at the University of Wisconsin-Madison, where she co-founded the Wisconsin Network for Research Support (WINRS), a nationally-recognized model for effective inclusion of diverse stakeholder perspectives to address health equity and advance translational science.
  - In her role at the UW-Madison, Gay developed an innovative service for researchers, called Community Advisors on Research Design and Strategies (or the CARDS®. This unique service has consulted with over 200 diverse health science research teams since late 2010. ***A session with the CARDS® is part of this workshop series.***
  - Gay has consulted on 12 PCORI-funded projects and with more than 100 health science research teams in academia, the VA, state/local government and the private sector. She brings that critical "10,000 hours of practice" in engagement work.
  - Gay will lead this workshop series and continue to work with CAIRIBU for the next several years.

Stakeholder Engagement Basics for Health Science Researchers: Putting the Pieces Together

SERIES AGENDA

1. **March 8**
   - Introductions, learning objectives for series, preview of coming sessions
   - Identify range of potential stakeholders (who)
   - Discuss the benefits of stakeholder engagement (why)
   - Homework (complete before session date):
     - Read: *Practical Guidance for Involving Stakeholders in Health Research*

2. **April12**
   - Review Best Practices for interacting with stakeholders and apply these practices to prepare strategic agenda for May CARDS® meeting (how)
   - Homework (complete before session date):
     - Read: *A Case Study of Engaging Hard-to-Reach Participants in the Research Process: Community Advisors on Research Design and Strategies (CARDS)®* .. p. 22
     - Read: *Power of the Personal* .. p. 31
     - Review: Sample stakeholder meeting agenda .. p. 35
     - Watch: Videos on CARDS;® link **HERE**

3. **May 10**
   - Participate in CARDS® meeting
   - Homework:
     - Before: Download, review How To Recruit and Retain Patient Stakeholders (HARPS) – watch select video clips
     - After: Complete short Qualtrics survey about CARDS® experience *(link to be provided)*

4. **June 14**
   Debrief as a group, review CARDS® summary, discuss HARPS/stakeholder engagement plans, get feedback from group on questions about stakeholder engagement plan development, discuss options for next steps (possibly continue as ongoing Research Interest Group, identify topics/issues for a few additional workshops, plan an additional CARDS® meeting, etc.)
   - Homework (complete before session date):
     - Continue to review HARPS; discuss application to developing an engagement plan for grant submission
     - Other resources ***(links to all are available on page 40)***:
       1. PAT-1
       2. PCORI online guides
       3. *Strengthening diversity in research partnerships: Knowledge to action guide*, Institute for Patient-and Family-Centered Care
       4. *Choosing the "Right" Patients to Avoid Pitfalls,* Hastings Institute

CAIRIBU Stakeholder Engagement Workshop Series - Cohort 1 (March-June 2022)

#	First and Last Name	Stage	Institutional Affiliation	CAIRIBU Affiliation	Type of Research	Research Focus	Prior Stakeholder Engagement Experience?	Stage of Research (e.g. preparing for research, conducting research, using research)	Stakeholder engagement plans — In what ways do you intend to be involved in stakeholder engagement in the future?	Which stakeholders are you most interested to include in your research?
1	David Bayne	ESI	UCSF	Current K12 Scholar (UCSF-Kaiser KUroEpi)	Clinical, Population Health	Access to surgical care for kidney stone disease.	N	Conducting research	Involve patients in qualitative research and gain input in how access to surgical care can be improved	Patients and the public; providers
2	Catherine Brownstein	Estab	Harvard Univ	Opportunity Pool awardee, Columbia Univ U54 O'Brien Center	Basic, Clinical, Translational		Y			
3	Christi Butler	Estab	UCSF	---	Clinical, Population Health	Population of interest is transgender and gender diverse individuals. research focus on those presenting for gender affirming surgery: motivation, outcomes	N	Preparing for research	As a provider of gender affirming surgery I plan to invest as a resource for counseling process, surgical technique, outcomes, and complications	Patients and public; providers; payers - many surgeries and pre surgical requirements not covered; policy makers; principal investigators
4	Jon Ellison	ESI	Children's Hospital of Milwaukee/ MCW	Investigator, MCW P20 Center	Clinical, Translational	Development of a patient-prioritized research agenda	Y	Elicitation of research themes	Development of a Kidney Stone Engagement Core (complete), elicitation (ongoing) - refinement - prrioritization of research agenda items	Patients, caregivers, researchers, and clinicians
5	Sonia Fargue	ESI	Univ Alabama Birmingham	Investigator, UAB P20 Center	Basic, Translational	Kidney stones, oxalate metabolism	N	Conducting research	Not sure. I realize the importance of it and need to gather information and examples to be able to formulate ideas.	PI, patients/public, providers
6	James Hokanson	ESI	MCW/ Marquette Univ	Prior K12 Scholar (Duke KURe); current investigator MCW P20 Center	Basic, Clinical, Translational	My research focus has two broad areas. First, the design of novel therapies, particularly with a focus on electrical stimulation, for urological dysfunction. Second is physiological testing and diagnostics, along with predictive modeling, with a focus on understanding which patients respond to which therapies. I am currently focused primarily on urgency urinary incontinence, with some neurogenic bladder work as well, although that may change in the future.	N	I am currently conducting funded research while also preparing for future research (grant prep)	I have no idea but in general it sounds important!!	Tough to choose, but I would probably say patients and the public, followed by policy makers, followed by providers, followed by everyone else
7	Maryellen Kelly	ESI	Duke Univ	Prior K12 Scholar (Duke KURe)	Clinical	Pediatric urology lower urinary tract symptoms and neurogenic bladder/bowel	Y	Conducting research currently and also preparing grants for future research	As a primary investigator I will be designing research studies which involve stakeholder engagement. I have applied for a PCORI recently that had a rather robust plan but need advice particularly on stakeholder engagement plans for smaller pilot projects.	Patients, providers, policy makers, hospital administrators, industry, public
8	Thomas Peterson	Grad student	UW-Madison	Trainee, UW-Madison U54 O'Brien Center	Translational	Role of Aryl Hydrocarbon Receptor In Lower Urinary Tract Dysfunction Etiology.	N	Conducting Research	I plan on becoming a researcher in clinical and pre-clinical studies after graduating. The stakeholders that I plan on engaging with are the public, policy makers, product makers, and principal investigators while conducting research in pre-clinical studies. While conducting research in clinical studies, I plan on interacting with principle investigators, the public, providers, payers, patients, and policy makers.	Currently, the stakeholders most interested in my research are principle investigators.
9	Nazema Siddiqui	ESI	Duke Univ	Prior K12 Scholar (Duke KURe)						
10	Ruchika Talwar	ESI	Univ Pennsylvania	---		Quality of life, implementation science, health policy		Preparing for research, conducting research	Creating research models to use to incorporate patient reported outcomes into bundled payment model structures. what domains actually matter to patients?	Patients/public, providers, payers, policy makers
11	Kristina Warner	ESI	UW-Madison	---	Clinical, Translational, Population Health	Examining health disparities within the field of urogynecology with the intent of learning more from communities and improving access to care, access to care, inclusion in population based studies, and treatment.	N	Preparing for qualitative study after reviewing and analyzing previously collected survey data	It is my intent to align myself with community organizations where I am able to explain my study question and how study data can benefit community members.	- Patients and public - Providers
12	Bernadette Zwaans	ESI	Beaumont Health/ Oakland Univ	PI, Beaumont Health-Oakland Univ P20 Center	Clinical, Translational	Benign urology including radiation cystitis, overactive bladder and interstitial cystitis	Y	Preparing for research	We had applied for a PCORI research grant which was the first grant that requested a research engagement plan. This was very new to me and thus I'd like to get more experience in how best to develop such a plan.	Patients and consumers; Clinicians; Healthcare providers

CAIRIBU Stakeholder Engagement Workshop Series - Cohort Contact Information

David Bayne	David.Bayne@ucsf.edu
Catherine Brownstein	Catherine.Brownstein@childrens.harvard.edu
Christi Butler	christi.butler@ucsf.edu
Jon Ellison	JEllison@chw.org
Sonia Fargue	sfargue@uab.edu
James Hokanson	jhokanson@mcw.edu
Maryellen Kelly	Maryellen.kelly@duke.edu
Thomas Peterson	ntpeterson3@wisc.edu
Nazema Siddiqui	Nazema.siddiqui@duke.edu
Ruchika Talwar	ruchika.talwar@pennmedicine.UPenn.edu
Kristina Warner	KWarner@uwhealth.org
Bernadette Zwaans	bernadette.zwaans@beaumont.org

Gay Thomas, MA - Workshop Coordinator and Consultant	gay@grthomasadvisors.com
Kristina Penniston, PhD, RDN - PI, CAIRIBU Interactions Core	penn@urology.wisc.edu
Jennifer Allmaras, MPH - Research Specialist, CAIRIBU Interactions Core	allmaras@wisc.edu

Stakeholder Engagement Basics for Health Science Researchers:
Putting the Pieces Together

SESSION 1

Stakeholder Engagement Basics for Health Science Researchers: Putting the Pieces Together

Please return this completed form to cairibu@urology.wisc.edu by Friday March 4 at 5 PM eastern time.

Name	
Research Focus	
Stage of Research (e.g. preparing for research, conducting research, using research)	
Stakeholder engagement plans -- In what ways do you intend to be involved in stakeholder engagement in the future?	
Using Concannon et al. 2018 as a guide, list which stakeholder groups you are most interested in involving in your research.	

CAIRIBU

Practical Guidance for Involving Stakeholders in Health Research

Thomas W. Concannon, PhD[1,2], Sean Grant, PhD[3], Vivian Welch, PhD MSc[4], Jennifer Petkovic, PhD[4], Joseph Selby, MD MPH[5], Sally Crowe, PG Dip[6], Anneliese Synnot, MPH[7,8], Regina Greer-Smith, MPH[9], Evan Mayo-Wilson, DPhil[10], Ellen Tambor, MA[11], and Peter Tugwell, MD MsC[12] for the Multi Stakeholder Engagement (MuSE) Consortium

[1]The RAND Corporation, Boston, MA, USA; [2]Tufts Clinical and Translational Science Institute, Tufts University, Boston, MA, USA; [3]The RAND Corporation, Santa Monica, CA, USA; [4]University of Ottawa Centre for Global Health, Ottawa, ON, Canada; [5]Patient Centered Outcomes Research Institute, Washington, DC, USA; [6]Crowe Associates Ltd, Oxford, UK; [7]School of Psychology and Public Health, La Trobe University, Melbourne, Australia; [8]Cochrane Australia, School of Public Health and Preventive Medicine, Monash University, Melbourne, Australia; [9]Healthcare Research Associates LLC, Chicago, IL, USA; [10]Johns Hopkins Bloomberg School of Public Health, Baltimore, MD, USA; [11]Center for Medical Technology Policy, Baltimore, MD, USA; [12]Department of Medicine and School of Epidemiology and Public Health, University of Ottawa, Ottawa Hospital Research Institute, Bruyere Research Institute, Ottawa, ON, Canada.

Stakeholder engagement is increasingly common in health research, with protocols for engaging multiple stakeholder groups becoming normative in patient-centered outcomes research. Previous work has focused on identifying relevant stakeholder groups with whom to work and on working with stakeholders in evidence implementation. This paper draws on the expertise of a team from four countries—Canada, Australia, the UK, and the USA—to provide researchers with practical guidance for carrying out multi-stakeholder–engaged projects: we present a list of questions to assist in selecting appropriate roles and modes of engagement; we introduce a matrix to help summarize engagement activities; and we provide a list of online resources. This guidance, matrix, and list of resources can assist researchers to consider more systematically which stakeholder groups to involve, in what study roles, and by what modes of engagement. By documenting how stakeholders are paired up with specific roles, the matrix also provides a potential structure for evaluating the impact of stakeholder engagement.

KEY WORDS: stakeholder engagement; patient engagement; patient-centered outcomes research; research design; international health.

J Gen Intern Med 34(3):458–63
DOI: 10.1007/s11606-018-4738-6
© Society of General Internal Medicine 2018

BACKGROUND

Stakeholder engagement in health research has become increasingly common as investigators, journal editors, and funders recognize its potential influence on the evidence we produce.[1, 2] With the expansion in recent years of patient-oriented and translational research, engagement of stakeholders—patients, clinicians, policy makers, and others, each including multiple members—is increasingly expected.[3–5] However, despite a growing number of completed and ongoing stakeholder-engaged research projects,[6–12] little is known about the quality of stakeholder engagement work, or its impact on evidence.[13]

Experience shows that researchers have questions about engaging with stakeholders: what is it, why is it important, who should be involved, how should they be involved, when in the research process should they be involved, and what difference will it make?[14] To address these questions, published frameworks describe how to identify stakeholders and how to synthesize the views of multiple groups.[15–24] Previous work also characterizes stages of research in which stakeholders can be engaged, such as involvement in prioritizing research questions before a study is underway, or developing robust dissemination and implementation plans once it is completed.[16, 20, 21, 24]

In this paper, we synthesize expert opinion and research resources from four countries into practical guidance on carrying out the activities that are needed between stakeholder identification and evidence implementation: what are the roles (i.e., what research activities can stakeholders be involved in) and modes (i.e., how do stakeholders and researchers interact?) by which researchers and stakeholders can work together, and how can a study team select from among the many options? To find answers, we formed a consortium—dubbed Multi Stakeholder Engagement or MuSE—of individuals from Canada, Australia, the UK, and the USA who represent a variety of viewpoints about research in health. Our consortium identified several challenges for stakeholder-engaged research and formed a writing team and review panel (Online Appendix A) composed of individuals to address this particular need (Online Appendix B for a summary of MuSE projects).

Our primary audience is health researchers who are embarking on stakeholder-engaged work. The paper will

Electronic supplementary material *The online version of this article (https://doi.org/10.1007/s11606-018-4738-6) contains supplementary material, which is available to authorized users.*

Received March 23, 2018
Revised August 30, 2018
Accepted October 25, 2018
Published online December 18, 2018

be helpful especially to students and early career investigators who want to build a portfolio of stakeholder-engaged research. It will also be helpful to experienced scientists who recognize the need to involve stakeholders in their work. Finally, it should be useful to researchers who are already working with stakeholders but need a concise resource for designing future studies. Non-researchers are an important secondary audience; this work might be adapted specifically for individual stakeholder communities.

We define "stakeholder" as an individual or group who is responsible for or affected by health- and healthcare-related decisions. We use the term stakeholder engagement when describing activities the researcher can take in this relationship and stakeholder involvement when describing the activities that either researchers or stakeholders can take. In this paper, the term roles is used to describe the research activities in which stakeholders may be involved. The term modes of engagement is used to describe the processes by which researchers and stakeholders interact with each other. Finally, engaging with stakeholders is not the same as studying their views. This paper addresses involvement of stakeholders as partners in, not as, subjects of research.

CONSIDERATIONS BEFORE EMBARKING ON STAKEHOLDER-ENGAGED RESEARCH

When it comes to developing an engagement plan, the study team faces a bewildering array of options: How will we build relationships and trust with stakeholders before the work begins? When should the activities start and finish and how does uncertainty about funding for the work play into the plan? Will contact be sustained in person or by phone, email, web, or other tele-communications? How frequently should the research team and stakeholders be in contact with each other? Will contacts be made individually or in groups? Will individuals representing different stakeholder communities work separately or in mixed groups? Will opinions be gathered in structured discussions, formal interviews, or a survey? Are consensus techniques needed? Taken together, these and other considerations make developing an engagement plan a daunting task, especially for those new to multi-stakeholder engagement.

These decisions may be easier to tackle in steps. Before diving into the details, it will help to reflect on why to involve stakeholders, who should be involved, and how extensively they may be involved. Once a team has articulated these aspects of the plan, the specific roles and modes of involvement may become clearer. We discuss these four constructs—the rationale, extent, roles, and modes of involving stakeholders—in more detail below. Text box 1 presents this material as a series of questions.

Text Box 1. Considerations before embarking on stakeholder-engaged research

What is the rationale for engaging stakeholders?
- What are the intrinsic reasons for working with stakeholders?
- How do you expect working with stakeholders can improve your research?
- How to you expect working with stakeholders will improve relevant health care or outcomes?

Which stakeholder communities will be engaged?
- What model will you use to identify relevant stakeholders (Online Appendix B)?
- Which of the stakeholder communities in your framework make decisions the research is meant to inform?
- Which stakeholder communities are affected by decisions the research is meant to inform?
- What are the preferences of stakeholder communities for how they wish to be engaged?

How extensively will the stakeholders be engaged?
- How will stakeholders be involved in preparing for research?
- How will stakeholders be involved in conducting the research?
- How will stakeholders be involved in using the research?
- How intensively can stakeholders be involved in each activity?
- What resources and time that can be devoted to engagement activities?

What are the appropriate roles and modes by which stakeholders may be engaged?
- Will stakeholders have control over the course of the project?
- Will stakeholders help the research team carry out the research?
- Will stakeholder provide input but neither direct nor help with the research directly?
- Will activities be conducted in person or remotely?
- Will activities be conducted with individuals?
- Will activities be conducted with groups?
- Will stakeholder communities be mixed in multi-stakeholder activities?

What conflict of interest procedures and conflict management resources are needed?

What Is the Rationale for Engaging Stakeholders?

Pre-specifying the rationale—or desired outcomes—of engagement is a critical factor in developing an engagement plan. By establishing the desired outcomes in advance, furthermore, the study team can later evaluate the extent to which expectations have been met. This section points at ways to characterize the expected value of stakeholder engagement.

To articulate the rationale for engagement, the team might consider both its intrinsic and instrumental imperatives. Intrinsic imperatives suggest that engagement is an end in itself. In other words, involving stakeholders may simply be the right thing to do, especially if public dollars are used. The principles of engagement call researchers and stakeholders to pursue intrinsic goods like autonomy, dignity, equity, inclusiveness, partnership, and participation[25, 26]; in contrast, superficial involvement can be insulting to stakeholders.[26] Instrumental imperatives suggest that engagement produces some other good worth having. For instance, involving stakeholders may make study questions more relevant, methods and approaches more transparent, findings more useful, and evidence more likely to be used in practice.[16] If researchers wish our work to have detectable impact on health-related decisions, we should involve decision makers as we carry the work out.

Whether involving stakeholders meets intrinsic and instrumental aims can be assessed through quantitative and qualitative evaluation. Do stakeholders report a sense of autonomy, dignity, self-determination, equity, inclusiveness, partnership, and participation? To what extent do stakeholders use the research results in decision-making? The ultimate goal of health research is to improve health at affordable costs. For this reason, it may be productive to map explicitly how stakeholder involvement is expected to lead to improved research outcomes, without exceeding a desired budget.

Which Stakeholder Groups Will Be Involved?

Previous frameworks have sought to help researchers identify stakeholders in their work. Concannon et al. identified seven types of stakeholders in the Tufts-RAND 7Ps taxonomy for engagement[16]; Deverka et al. identified eight types in the Center for Medical Technology Policy framework for engagement[17]; PCORI identified nine types in its engagement "rubric."[19] Cochrane identified four audiences in its recent knowledge translation strategy.[20] Tugwell et al. named six types in a WHO Bulletin on knowledge translation for systematic reviews.[21] By putting these models in a single table (Online Appendix C), it is possible to see that differences in the *number of* stakeholder types are largely a matter of classification, not a disagreement about who should be included. At least two of the approaches[16, 17] were developed and published simultaneously, suggesting independent agreement about which stakeholders are key to improving health research.

Each of these frameworks recognizes that identifying the right individuals to represent stakeholder perspectives is a challenge, as stakeholders within a single group may hold different views. Choosing a multi-stakeholder approach is even more complex given the need to consider how to assure that underrepresented voices are heard, to ensure that the financial or academic interests of one group do not dominate the discussion, to manage group interactions and potential power imbalances, and to synthesize the views of different groups. It is important to be transparent about who was involved and why.

How Extensively Will Stakeholders Be Involved?

The frameworks described above agree that stakeholders can be involved throughout the research process and that contact should be sustained over time. The PCORI model identifies nine distinct research activities in three groups: (1) planning the study, (2) conducting the study, (3) disseminating the study results. Similarly, the Tufts-RAND 7Ps model lumps research activities into three stages: preparing for, conducting, and using research. The Cochrane model describes stakeholder involvement in the topic and question selection, design, execution, interpretation, and dissemination of their research.

When preparing for a study, researchers may engage stakeholders in a variety of activities, such as capacity building, team building, training, and topic selection. During the conduct of a study, activities like question development, selection of outcomes, participant recruitment, data collection, analysis, and identification and interpretation of findings may involve stakeholders. Once findings have been established and a study is concluded, researchers are often engaged in helping decision-makers use their findings. Stakeholders can be valuable partners during this stage by supporting or leading implementation activities.

Researchers may also consider how intensive the engagement of stakeholders should be. Some partner with stakeholders as co-investigators, sharing full control over the direction, management, and budget of a study. Others may arrange for stakeholder involvement at the level of technical advice. Thus, stakeholders may be empowered with differing levels of control over a study, from providing direction to collaborating, consulting, and providing information. It is possible, however, to mix approaches over the course of one or more studies and across different stakeholder groups.

What Are the Appropriate Roles and Modes by Which Stakeholders May Be Involved?

The core challenges we address include how to identify the roles that stakeholders will have and the modes by which their involvement will be facilitated. In designing the roles and modes of engagement, research teams should be responsive to stakeholder views about how they would like to be involved.[27–31] Researchers might try to learn from stakeholders about their interest in the research topic, including what the stakeholders want to get out of being involved. Furthermore, researchers should consider communicating about the interests and potential roles of stakeholders before a plan is settled. Once the plan is drafted, study teams may find that summarizing the engagement plan in a written document will facilitate communication. One recent work suggested summarizing roles and responsibilities in a "terms of reference" document for use by the whole research team throughout the project.[32]

Roles. All study teams—even those in basic and clinical sciences—have experience working with independent peers who review study protocols and manuscripts. This is a form of stakeholder engagement, in which external researchers with an interest in safeguarding the ethical conduct and rigor of research use commonly held standards to review the proposed or completed work.

Engagement with non-research stakeholders is similar. This might involve assembling a panel of individuals who have an interest in the outcomes of the research and can potentially use it to support decisions. Expert panels can review documents, run practice tests of survey instruments, vote on the relevance or importance of evidence for decisions they make, or work together to identify the implications of study results for their communities. They can develop study inclusion criteria, discuss and revise study protocols and materials, and identify outcomes that are most important to them.[33, 34]

Engagement activities may not be oriented toward finding a group's consensus about research, but rather on identifying the way that different stakeholder groups view the work. Understanding whether and how stakeholders disagree about the research can be just as important as understanding whether and how they agree. Knowing about these views may be critical to public release of the evidence with appropriate messaging and context.

Investigators working in community-based participatory research (CBPR), partnered action research, and co-produced research have experience partnering with communities in the direction, management, and oversight of studies. Involvement in this work may include deciding which work to prioritize, setting or sharing budgets, and managing personnel. It is critical to remember that principal investigators—whether researchers or stakeholders—retain full responsibility for the ethical conduct, quality, and rigor of research. The process of pre-publication review by scientific peers safeguards these dimensions of quality, and these reviews may be conducted transparently in the presence of stakeholders.

Modes. Distinct from the roles stakeholders play are the modes of interaction. While roles refer to the activities of stakeholders as they become involved in the work, modes have to do with the format and structure of interactions between researchers and stakeholders. When stakeholders are invited to serve as co-investigators or collaborators, the modes of engagement typically include all of the routine communication channels and interaction opportunities of the research workplace, such as in-person meetings, chance meetings, telephone calls, e-mails, and web-enabled communications. In instances where stakeholders serve in a consulting or advisory role, the modes of interaction may involve specialized communications in person, by telephone, by e-mail, or over the web. Alternatively, modes of interaction may include group communications such as town meetings, or group discussions, and these can also be held in-person or virtually. Finally, passive modes of communication are also used, such as public comment periods for research prioritizations or plans, and comments may be collected in writing, electronically, or by telephone.

WRITING AN ENGAGEMENT PLAN

Once these considerations have been reviewed, researchers and stakeholders can work together to write an engagement plan. To assist, we introduce a matrix that may be used to summarize a stakeholder engagement approach for a program of research or a single study (Table 1).

The rows describe stages and illustrative activities in a research project. While the rows may imply a sequential process from top to bottom, in practice, studies involve several iterative steps that loop backward and jump forward. The columns describe stakeholder groups, derived from a combination of the frameworks[16, 17, 19–21] (Online Appendix C). The collection of activities and stakeholders might be described at length in a legend. Researchers can use the matrix to develop a plan, share and revise the plan after consultation with stakeholders about how they would like to be involved, or summarize completed work involving stakeholders of different types in activities of different types. The table facilitates critical appraisal of the engagement plan by providing a quick view over the whole project. Researchers may decide to publish a completed matrix in their proposals or manuscripts.

Each cell in the matrix may be filled in with information summarizing the roles and modes of engagement for a specific stakeholder group in the research activity. Filling in many or most cells in this matrix will make the table quite large. As the table grows in size, it will become clear that the time and resource costs of engaging with stakeholders are significant. It also serves to point out where there are gaps in the plan.

This matrix is meant to help in planning well-targeted engagement activities. It is not meant to imply that all research activities require engagement, nor that all stakeholder groups must be included in all activities. There may be good reasons why certain cells are left blank; in these cases, researchers might note the rationale for blank cells or gaps in the matrix. In many cases, the roles and modes of involvement will be identical for stakeholders representing different communities. For instance, a technical advisory board composed of patients, clinicians, payers, and policy makers might be charged with several activities: (1) reviewing the study design; (2) reviewing data analyses; and (3) participating in identifying and interpreting findings.

Finally, any stakeholder who represents one community may belong to one or more additional communities. For example, some purchasers are also payers and some payers provide care. Patients and their advocates may also be providers or employers with policy-making responsibilities. Overlap may be inevitable, but care should be taken that multiple roles do not create unacceptable conflicts of interest. Building relationships with stakeholders, addressing conflict of interest, and dealing with conflict between those with competing interests are topics that have been explored elsewhere. Research teams need formal processes to build relationships and address conflicts, and resources are available to help.[35–38]

DISCUSSION

We developed this guidance to assist researchers to choose appropriate roles and modes of engagement with multiple stakeholder communities. Stakeholder engagement is in need of further experimentation, and the exact path toward meaningful engagement will not be uniform across every research institution and project. Our recommendations for developing stakeholder engagement plans follow a plan-do-study-act (PDSA) approach, and they can be applied to any program:

Table 1 Summarizing a stakeholder engagement plan

Research stage	Research activity	Stakeholder group						
		Patients and the public	Providers	Payers	Purchasers	Product makers	Policy makers	Principal investigators
Preparing for research	Building research capacity of stakeholders							
	Training researchers to work with stakeholders							
	Prioritizing evidence gaps							
	Choosing research topics							
Conducting research	Defining the research question							
	Choosing relevant outcomes							
	Designing a research protocol							
	Defining participant inclusion and exclusion criteria							
	Drafting or revising study materials and protocols							
	Recruiting participants							
	Monitoring patient data and safety							
	Collecting data							
	Analyzing data							
	Identifying findings							
	Interpreting findings							
	Disseminating results							
Using research	Implementing evidence in practice							
	Evaluating research							
	Evaluating engagement							
	Identifying topics for future research							

1. *Set forth engagement roles and modes (plan).* Researchers should plan engagement roles and activities throughout the course of a study, and funders might incorporate expectations about doing so in funding opportunity announcements.
2. *Experiment with alternative strategies (do).* Research institutions, investigators, and stakeholder groups should begin to experiment with organizational- and project-level strategies to support stakeholder engagement. Institutions might initiate stakeholder activities to support multiple studies, such as a priority-setting process to inform the organization's research agenda. Investigators can begin to experiment with alternative approaches to engagement in their own research.
3. *Evaluate alternative approaches (study).* Funders and investigators can begin right away to identify appropriate intermediate and long-term benchmarks for evaluating the effectiveness of roles and modes, keeping in mind that the optimal approach will vary by institution and project. Future research on what works and what does not might consider whether and what kind of stakeholder engagement leads to informed decision-making and improved uptake of evidence into practice. The matrix offered in Table 1 can help with this challenge by pointing at specific activities we hope to change (in rows) and how we hope to change them (in cells). The resources listed in Online Appendix D can help researchers adapt their approach to different contexts.
4. *Report on outcomes, implement changes as needed, and iterate (act).* Investigators should consider reporting stakeholder activities in manuscripts and contract reports. Journals might consider publishing both quantitative and qualitative research on this topic, to continue establishing an evidence base about what works in various settings. As the evidence base grows, funders, research institutions, and investigators need to be prepared to implement changes in their engagement programs. As changes are adopted, an iterative assessment process should follow.

In this paper, we have offered practical guidance on designing and carrying out an engagement plan. Research teams and funders may use pre-specified aims and roles and modes of engagement to support formal evaluations of stakeholder engagement work, an important next step.

Acknowledgements:

Contributors: *The authors wish to thank Amanda Borsky, Angela Coulter, Zoë Gray, Jeanne-Marie Guise, Sophie Hill, Joan Powell, Laurel Pracht, Beverly Rogers, and Beverly Shea for sharing resources from a variety of countries and for their detailed reviews of an earlier version of this manuscript. We thank MuSE project manager Jennifer Vincent for*

keeping this work on track and RAND colleague Kristin Sereyko for preparing the manuscript for publication.

Prior Presentations: *This paper has not been presented at any previous conferences.*

Corresponding Author: *Thomas W. Concannon, PhD; The RAND Corporation, Boston, MA, USA (e-mail: tconcann@rand.org).*

Compliance with Ethical Standards:

Conflict of Interest: *RG-S is President of Healthcare Research Associates, a for-profit consultancy based in Chicago, IL, that specializes in patient engagement in research. RG-S holds a contract with Boehringer Ingelheim. SC is a principal with Crowe Associates Limited, a for-profit company registered in England and Wales that specializes in patient and public involvement in research. SG's spouse is a salaried employee of Eli Lilly and Company; SG owns stock in the company and has accompanied his spouse on company-sponsored travel. All other authors declare no conflicts of interest specific to this manuscript.*

Publisher's note *Springer Nature remains neutralwith regard to jurisdictional claims in published maps and institutional affiliations.*

REFERENCES

1. **Frank L**, **Basch E**, **Selby JV**. The PCORI perspective on patient-centered outcomes research. JAMA 2014;312(15):1513–1514.
2. **Viswanathan M**, **Ammerman A**, **Eng E**, **et al.** Community-Based Participatory Research: Assessing the Evidence. Evidence Report/Technology Assessment No. 99. AHRQ Publication 04-E022. Rockville, MD: Agency for Healthcare Research and Quality; 2004.
3. Patient-Centered Outcomes Research Institute. What we mean by engagement. Available at: https://www.pcori.org/engagement/what-we-mean-engagement. Accessed 24 Sept 2018.
4. **Cottrell E**, **Whitlock E**, **Kato E**, **et al.** Defining the benefits of stakeholder engagement in systematic reviews. Research White Paper. AHRQ Publication No. 14-EHC006-EF. Rockville, MD: Agency for Healthcare Research and Quality; 2014.
5. INVOLVE. Briefing notes for researchers: involving the public in NHS, public health and social care research. Eastleigh, UK: INVOLVE; 2012.
6. **Domecq JP**, **Prutsky G**, **Elraiyah T**, **et al.** Patient engagement in research: a systematic review. BMC Health Serv Res 2014;14(1):1.
7. **Concannon TW**, **Fuster M**, **Saunders T**, **et al.** A systematic review of stakeholder engagement in comparative effectiveness and patient-centered outcomes research. J Gen Intern Med 2014;29(12):1692–1701.
8. **Shippee ND**, **Domecq Garces JP**, **Prutsky Lopez GJ**, **et al.** Patient and service user engagement in research: A systematic review and synthesized framework. Health Expect 2015;18(5):1151–1166.
9. **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 2016;31(1):13–21.
10. **Shea B**, **Santesso N**, **Qualman A**, **Heiberg T**, **Leong A**, **Judd M**, Consumer-driven health care: building partnerships in research. Health Expect 2005;8(4):352–359.
11. **de Wit M**, **Kirwan JR**, **Tugwell P**, **et al.**. Successful stepwise development of patient research partnership: 14 years' experience of actions and consequences in outcome measures in rheumatology (OMERACT). Patient 2017;10(2):141–152.
12. **Urquhart-Secord R**, **Craig JC**, **Hemmelgarn B**, **et al.**. Patient and caregiver priorities for outcomes in hemodialysis: an international nominal group technique study**.** Am J Kidney Dis 2016; 68(3):444–458.
13. **Esmail L**, **Moore E**, **Rein A**. Evaluating patients and stakeholder engagement in research: moving from theory to practice. J Comp Eff Res 2015;4(2):133–145.
14. **Forsythe LP**, **Frank LB**, **Workman TA**, **et al.**. Health researcher views on comparative effectiveness research and research engagement. J Comp Eff Res. 2017;6(3):245–56. Available at: https://doi.org/10.2217/cer-2016-0063. Accessed 24 Sept 2018.
15. **Mullins CD**, **Abdulhalim AM**, **Lavallee DC**. Continuous patient engagement in comparative effectiveness research. JAMA 2012;307(15):1587–1588.
16. **Concannon TW**, **Meissner P**, **Grunbaum JA**, **et al.** A new taxonomy for stakeholder engagement in patient-centered outcomes research. J Gen Intern Med 2012;27(8):985–991.
17. **Deverka PA**, **Lavallee DC**, **Desai PJ**, **et al.** Stakeholder participation in comparative effectiveness research: Defining a framework for effective engagement. J Comp Eff Res 2012;1(2):181–194.
18. **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 2015;24(5):1033–1041.
19. Patient-Centered Outcomes Research Institute. PCORI's Stakeholders. Available at: http://www.pcori.org/funding-opportunities/what-we-mean-engagement/pcoris-stakeholders. Accessed 24 Sept 2018.
20. The Cochrane Collaboration. Cochrane Knowledge Translation Strategy. Available at: http://community.cochrane.org/sites/default/files/uploads/inline-files/Cochrane%20Knowledge%20Translation%20Strategy%20FINAL%20for%20-website.pdf, April 2017. Accessed 24 Sept 2018.
21. **Tugwell P**, **Robinson V**, **Grimshaw J**, **Santesso N**. Systematic reviews and knowledge translation. Bull World Health Organ 2006;84(8):643–51.
22. **Schiller C**, **Winters M**, **Hanson HM**, **Ashe MC**. A framework for stakeholder identification in concept mapping and health research: a novel process and its application to older adult mobility and the built environment. BMC Public Health 2013;13:428.
23. **O'Haire C**, **McPheeters M**, **Nakamoto EK**, **et al.** Methods for engaging stakeholders to identify and prioritize future research needs. Methods Future Research Needs Report No. 4. AHRQ Publication No. 11-EHC044-EF. Rockville, MD: Agency for Healthcare Research and Quality; 2011.
24. **Esposito D**, **Heeringa J**, **Bradley K**, **Croake S**, **Kimmey L**. PCORI Dissemination and Implementation Framework. Princeton, NJ: Mathematica Policy Research; 2015.
25. **Morain SR**, **Concannon TW**, **Wittenberg E**. HIV, breast cancer and vaccines: what to high-profile cases reveal about stakeholder engagement in research? J Comp Eff Res 2017;6(1):43–49.
26. **Gradinger F**, **Britten N**, **Wyatt K**, et al.. Values associated with public involvement in health and social care research: a narrative review. Health Expect 2015;18(5):661–675.
27. **Concannon TW**, **Kotzias V**, **Khodyakov D**, **Graff J**, **Fahey G**. What makes stakeholders want to become involved in research? The RAND Blog. 2016. Available at: https://www.rand.org/blog/2016/05/what-makes-stakeholders-want-to-become-involved-in.html. Accessed 24 Sept 2018.
28. **Concannon TW**, **Khodyakov D**, **Kotzias V**, **Fahey G**, **Graff J**, **Dubois RW**. Employer, Insurer, and Industry Perspectives on Patient-Centered Comparative Effectiveness Research. Santa Monica, CA: RAND Corporation, 2015. Available at: http://www.rand.org/pubs/research_reports/RR1242.html. Accessed 24 Sept 2018 .
29. **Concannon TW**, **Friedberg MW**, **Hwang A**, **Wiitala K**. Engaging Consumers in the Quality Measurement Enterprise. Santa Monica, CA: RAND Corporation, 2017. Available at: https://www.rand.org/pubs/research_reports/RR1760.html. Accessed 24 Sept 2018.
30. **Concannon TW**, **Kotzias V**, **Khodyakov D**, **Fahey G**, **Graff J**. What do stakeholders say about comparative effectivess research? *The RAND Blog*. 2016. Available at: https://www.rand.org/blog/2016/04/what-do-stakeholders-say-about-comparative-effectiveness.html. Accessed 24 Sept 2018.
31. **Forsythe LP**, **Frank LB**, **Workman TA**, **Hilliard T**, **Harwell D**, **Fayish L**. Patient, caregiver and clinician views on engagement in comparative effectiveness research. J Comp Eff Res 2017;6(3):231–44. Available at: https://www.futuremedicine.com/doi/full/10.2217/cer-2016-0062. Accessed 24 Sept 2018.
32. **Xian Y**, **Wu J**, **O'Brien EC**, et al.. Real world effectiveness of warfarin among ischemic stroke patients with atrial fibrillation: observational analysis from Patient-Centered Research into Outcomes Stroke Patients Prefer and Effectiveness Research (PROSPER) study. BMJ 2015;351:h3786. doi: https://doi.org/10.1136/bmj.h3786.indd.
33. **Concannon TW**. Can Patient-Centered Outcomes Research Improve Health Care? BMJ 2015;351:h3859.
34. **Jull J**, **Mazereeuw M**, **Sheppard A**, **Kewayosh A**, **Steiner R**, **Graham ID**. Tailoring and field-testing the use of a knowledge translation peer support shared decision making strategy with First Nations, Inuit and Métis people making decisions about their cancer care: a study protocol. Res Involv Engagem. 2018;4(6). DOI https://doi.org/10.1186/s40900-018-0085-3
35. Institute of Medicine. Conflict of Interest in Medical Research, Education, and Practice. Washington: National Academies Press; 2009.
36. Harvard Business School Press. The Essentials of Negotiation. Boston: Harvard Business School Publishing; 2005.
37. **Malhotra D**, **Bazerman MH**. Negotiation Genius. New York: Bantam Books; 2008.
38. Institute of Medicine. Clinical Practice Guidelines We Can Trust. Washington: The National Academies Press; 2011.

Session 1 Presentation - Gay Thomas

Getting started in Stakeholder Engagement:

Putting the pieces together

Spring 2022
4-part workshop series

CAIRIBU

1

GR THOMAS ADVISORS LLC
ENGAGING DIVERSE VOICES TO PROMOTE RESEARCH AND POSITIVE HEALTH OUTCOMES

Helpful tips for Virtual Meetings

- Find a quiet, private place to sit for the whole meeting.
- Log in 5-10 minutes before the start time
- Consider using headphones for better sound
- Stay muted when you aren't speaking.
- Use "raise hand" feature, literally raise your hand or use the chat box to let us know you've got a comment/question.
- Allow pauses after speaking so colleagues aren't talking over one another. (There can be small delays with video)
- When you talk, please start by saying "This is [your name]..... So we know who is speaking.
- Keep your video on during the meeting when possible.

2

Overview & Introductions

4-part workshop series

- Learning objectives
- Session preparation
- Overview of workshop sessions
- Questions?

Introductions:

- Name and one positive word a friend/relative would use to describe you

3

WHO are Stakeholders in Research?

- Concannon TW, Meissner P, Grunbaum JA, McElwee N, Guise JM, Santa J, Conway PH, Daudelin D, Morrato EH, Leslie LK. A new taxonomy for stakeholder engagement in patient centered outcomes research. Journal of General Internal Medicine, 2012 Aug; 27 (8):985-991. doi: 10.1007/s11606-012-2037-1

4

WHY involve Stakeholders?

"Projects are expected to involve collaborations with relevant organizations or groups or stakeholders, such as academic institutions, health service providers and systems, state and local public health agencies or other governmental agencies such as housing and transportation, criminal justice systems, school systems, patient or consumer advocacy groups, community-based organizations, and faith-based organizations."

https://www.niddk.nih.gov/research-funding/current-opportunities/rfa-md-21-004.

5

The Department of Defense Spinal Cord Injury Research Program (SCIRP)

Applicants are asked to involve people with an SCI, their family members, and/or their care partners to provide ongoing advice and consultation throughout the planning and implementation of the research project. The SCIRP believes that capturing and integrating the unique perspectives and experiences of these individuals will enable better and more impactful outcomes for people living with SCI.

6

Researchers benefit from an outside perspective

7

Stakeholder impact on research materials

Cognition and Heart Failure Study

- *Thinking and Memory: The Heart-Brain Connection*

Detection and Neurological Impact of Cerebrovascular Events In Noncardiac Surgery Patients

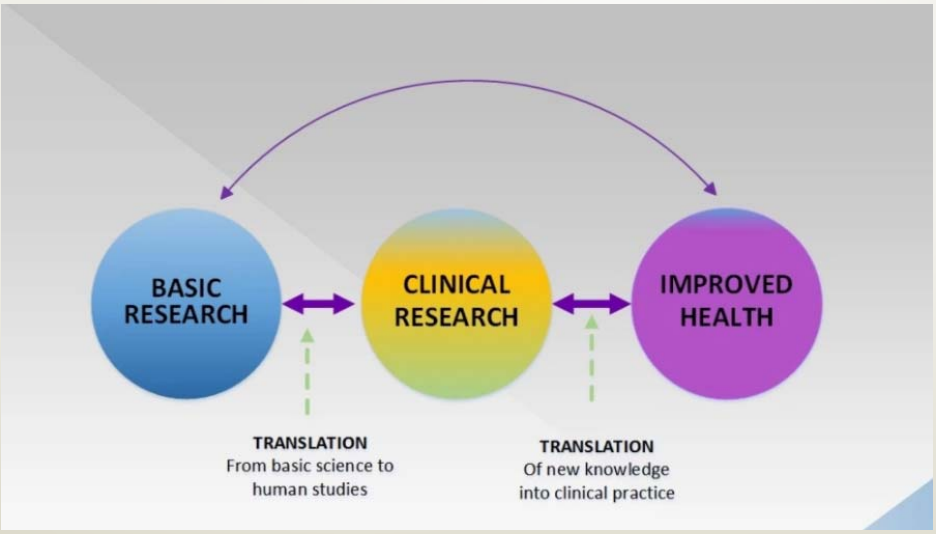
Learning more about strokes during surgery

Question:

Does your child live or stay in more than one home on a regular basis?

8

Connection to Basic Science



9

Image adapted from: Zerhouni EA. 2007. Translational research: moving discovery to practice. Clinical Pharmacology and Therapeutics 81:126-128.

Example from Rockefeller University

Used with permission of the Rockefeller University Center for Clinical Translational Science.

Patient-reported outcomes led to exploring new hypothesis

10

Benefits of Engagement

- Improve study design
- Improve data collection instruments and processes
- Help interpret study findings
- Improve recruitment
- Increase retention rates
- Help develop materials and processes for disseminating study results to various audiences
- Enhance communication with all stakeholders
- Improve public/community attitude toward research & researchers; build trust

11

Review – learning objectives

✓ Identify stakeholder categories and groups most relevant to research team
✓ Understand how stakeholders can affect research and research outcomes
✓ Understand how stakeholder engagement could add value to your program of research

- Describe key steps in planning stakeholder meetings
- Recognize important elements in a stakeholder engagement plan
- Gain awareness of existing resources to support stakeholder engagement

12

Preview of Coming Attractions

- Tues. April 12th - HOW of engagement
  - Focus on making effective use of time with stakeholders
  - Work together to prepare for May meeting with CARDS
- Background preparation to get the most of out April workshop:
  - Read: *A Case Study of Engaging Hard-to-Reach Participants in the Research Process: Community Advisors on Research Design and Strategies (CARDS)®*
  - Watch 1-3 brief videos on CARDS

13

GR THOMAS ADVISORS LLC
ENGAGING DIVERSE VOICES TO PROMOTE RESEARCH AND POSITIVE HEALTH OUTCOMES

NOTES FROM SESSION 1

Stakeholder Engagement Basics for Health Science Researchers:
Putting the Pieces Together

SESSION 2

SESSION 2, April 12

PRE-SESSION HOMEWORK:

1. Read Kaiser et al, ***"A case study of engaging hard-to-reach participants in the research process: community advisors on research design and strategies (CARDS)®"*** in *Research in Nursing & Health* 2016, pages 22-30 in this booklet
2. Read Thomas et al, ***"The power of the personal: breaking down stereotypes and building human connections"*** in *Native Inquiry in Bioethics* 2017, pages 31-34 in this booklet
3. Watch videos on the website of the Wisconsin Network for Research Support, ***HERE***
4. Review sample agenda for a stakeholder meeting, pages 35-36 in this booklet

A Case Study of Engaging Hard-to-Reach Participants in the Research Process: Community Advisors on Research Design and Strategies (CARDS)®

Betty L. Kaiser, Gay R. Thomas, Barbara J. Bowers

Correspondence to: Betty L. Kaiser
E-mail: blkaiser@wisc.edu

Betty L. Kaiser
Administrative Program Specialist
School of Nursing
University of Wisconsin-Madison
701 Highland Ave., Rm. 5138
Madison, WI 53705

Gay R. Thomas
Senior Administrative Program Specialist
School of Nursing
University of Wisconsin-Madison
Madison, WI

Barbara J. Bowers
Professor
School of Nursing
University of Wisconsin-Madison
Madison, WI

Abstract: Lack of diversity among study participants in clinical research limits progress in eliminating health disparities. The engagement of lay stakeholders, such as patient or community advisory boards (CABs), has the potential to increase recruitment and retention of underrepresented groups by providing a structure for gathering feedback on research plans and materials from this target population. However, many CABs intentionally recruit prominent stakeholders who are connected to or comfortable with research and academia and thus may not accurately represent the perspectives of underrepresented groups who have been labeled hard-to-reach, including racial minorities and low-income or low-literacy populations. We developed a partnership between the University of Wisconsin-Madison School of Nursing and two community centers to deliberately engage hard-to-reach people in two lay advisory groups, the Community Advisors on Research Design and Strategies (CARDS)®. Community center staff recruited the CARDS from center programs, including parenting and childcare programs, women's support groups, food pantries, and senior meal programs. The CARDS model differs from other CABs in its participants, processes, and outcomes. Since 2010, the CARDS have met monthly with nurses and other researchers, helping them understand how research processes and the language, tone, appearance, and organization of research materials can discourage people from enrolling in clinical studies. We have successfully used the CARDS model to bring hard-to-reach populations into the research process and have sustained their participation. The model represents a promising strategy for increasing the diversity of participants in clinical research. © 2016 Wiley Periodicals, Inc.

Keywords: subject recruitment; healthcare disparities; vulnerable populations; advisory committees; stakeholder engagement

Research in Nursing & Health
Accepted 19 August 2016
DOI: 10.1002/nur.21753
Published online in Wiley Online Library (wileyonlinelibrary.com).

The elimination of health disparities is a national priority (National Institute on Minority Health and Health Disparities, 2016; US Department of Health and Human Services, 2016). Clinical researchers can play an important role in eliminating health disparities by making novel treatments available to underserved populations and identifying the effectiveness of particular treatments for particular populations. However, progress toward eliminating health disparities has been hindered by lack of participant diversity in clinical research studies (Kitterman, Cheng, Dilts, & Orwoll, 2011; Schroen et al., 2010). Researchers often do not reach recruitment goals for hard-to-reach participants such as racial and ethnic minorities and people with low income. Reasons for low recruitment include mistrust of research, perceived risks, and lack of culturally appropriate information about opportunities to participate in research (Ford et al., 2008), as well as burdens such as time commitment and lengthy questionnaires (Paskett et al., 2008).

© 2016 Wiley Periodicals, Inc.

A recommended strategy for overcoming barriers to recruitment is patient and community stakeholder involvement in the design and conduct of clinical trials (Institute of Medicine, 2011). One popular form of stakeholder involvement is community advisory boards (CABs). CABs serve several functions, depending on the project mission and requirements. They provide input into research agendas; serve as gatekeepers for researcher entry into communities; formally approve projects; and influence the design, conduct, and implementation of studies (Fernandez-Pena et al., 2008; Silvestre, Quinn, & Rinaldo, 2010; Strauss et al., 2001).

Recommendations for CAB membership often emphasize inclusion of influential stakeholders and opinion leaders from the community (D'Alonzo, 2010). Typical CAB members include service providers, community leaders, and representatives of local agencies and organizations (Newman et al., 2011); in this paper, high-level stakeholders are referred to as "prominent community representatives." Prominent community representatives bring valuable skills, perspectives, and resources to CABs due to their experience with group and meeting processes, influence and connections within the community, and decision-making power within their organizations.

CABs also sometimes include lay stakeholders who are not prominent community representatives but can bring an important viewpoint to a research project, such as residents of a particular neighborhood or patients or caregivers with experience related to a particular health issue (James et al., 2011; Pinto, Spector, Rahman, & Gastolomendo, 2013). Lay stakeholders can help researchers improve the cultural sensitivity and appropriateness of recruitment materials and methods; recommend study implementation strategies (Joosten et al., 2015); and design relevant, meaningful interventions (GreenMills, Davison, Gordon, Kaigang, & Jurkowski, 2013). The perspectives of lay stakeholders who are disconnected from academia and local power structures may offer clinical researchers unique insights and perspectives on recruitment barriers and strategies for improving recruitment and retention of hard-to-reach populations. In this paper, we (the authors) describe the origins, participants, processes, and outcomes of a unique CAB comprised solely of lay stakeholders that brings voices of rarely heard groups into the research enterprise.

Project Origins and Community—Academic Partnership

Our team at the University of Wisconsin-Madison (UW-Madison) School of Nursing partnered with two local community centers to develop lay advisory boards of community members who are not prominent community representatives. The Lussier Community Education Center and Goodman Community Center have served their neighborhoods for over 30 years, offering programs and services that reflect the diversity of their communities. In 2010, we worked with staff at the community centers to submit a proposal for a program grant to the National Institutes of Health (NIH). We received a 3-year grant to establish a sustainable infrastructure to link community members with researchers to improve the quality of health sciences research. The UW-Madison Health Sciences Institutional Review Board (IRB) designated our grant activities as exempt from review.

In one of our grant activities, we worked with the community centers to develop and pilot two lay advisory boards, the Community Advisors on Research Design and Strategies (CARDS)®. We established a CARDS group at each partnering community center. For the past 5 years, each group has met monthly with guest researchers to provide feedback on the appeal, clarity, and accessibility of materials and processes used in clinical research. Our NIH grant ended in 2013, and at the time of this writing we are sustaining the CARDS as a program within the Wisconsin Network for Research Support (WINRS), a fee-for-service community and patient engagement center supported by the School of Nursing (http://winrs.son.wisc.edu/). The UW-Madison Institute for Clinical and Translational Research, a Clinical and Translational Sciences Award site, provided additional support for the program.

The CARDS® Program

The CARDS are based in the community, so researchers often assume that CARDS input is relevant only for researchers interested in community-based participatory research (CBPR) or community-engaged research (CER). In fact, since the program's inception, the CARDS have worked extensively with researchers conducting clinic or hospital-based studies. The CARDS provide lay, patient-centered feedback on materials for any type of human subjects research, regardless of the specific research topic, setting, methodology, or study population. To date, researchers and their project staff typically attended one or two meetings for each project that they brought to the CARDS, although we placed no limit on the number of CARDS meetings that a researcher may attend. The CARDS program makes it feasible for researchers who are not conducting CBPR or CER to get timely, meaningful lay input on their research materials, without substantial investment of resources.

Program Participants

Members. For the CARDS, we wanted not prominent community representatives, but people who have been underrepresented in healthcare research (Hasnain-Wynia & Beal, 2014). We intentionally sought to recruit people who are not connected with academia, research,

healthcare, or local power structures. Unlike recruitment for many CABs, we did not establish specific inclusion criteria, such as residency in a certain neighborhood or diagnosis of a particular health condition (Stewart et al., 2015). We asked community center staff (CARDS liaisons) to recruit typical users of services at their centers. Liaisons shared an informational flyer with community members who used center services such as parenting and childcare programs, women's support groups, job clinics, food pantries, and senior meal programs. People who were interested followed up with the CARDS liaison to complete a membership application, on which they were asked to briefly explain why they wanted to be part of the CARDS. Sometimes, people who worked or volunteered at the centers heard about the program and completed applications. At the time of this writing, 15 CARDS members were participating in the program. Nine participants (60%) were women and 10 (67%) were African-American. Ages ranged from early 20s to mid-70s, and educational attainment ranged from incomplete high school preparation to post-high-school coursework.

Many CABs have expansive roles for their members that may encompass defining research agendas, providing entrée into a community (gatekeeping), engaging in the conduct of research, and providing feedback on scientific papers (Israel et al., 2005). The sole function of the CARDS is to meet monthly with researchers to review, discuss, and provide feedback on research plans and materials, including recruitment materials, data collection procedures and instruments, web-based materials, and smartphone apps. CARDS members are never required to complete work between meetings. For each 90-minute meeting, they receive a cash stipend of $35. Although it is convenient for institutions to pay board members with checks, many people in hard-to-reach populations do not have checking accounts. As one of the CARDS explained, "$35 isn't $35 if I have to pay a fee to cash the check."

WINRS and community center staff. WINRS staff provided overall management of the CARDS program. The School of Nursing employed two WINRS staff members, at a total of 1.7 full-time equivalents. WINRS staff worked on a variety of projects but together devoted approximately 0.5 full-time equivalents to management of the CARDS. Both employees had advanced degrees (MA; PhD) and had previous experience with community engagement. One employee had primary responsibility for the program, with the other providing support. The School of Nursing and ICTR provided salary support for the full-time employee, and revenues from WINRS supported the salary of the part-time employee. In addition, each community center provided a staff member who served as the center's CARDS liaison. Table 1 lists the respective responsibilities of WINRS, community center staff, and the CARDS. We paid the centers a monthly facility fee to compensate them for liaison time, room rental, child care during meetings, and transportation for members as needed. Monthly facility fees ranged from $205 to $290, depending on whether child care was provided.

Guest researchers. Since the CARDS program began in 2010, we have conducted meetings with 40 research teams representing 21 disciplines, including cardiology, family medicine, and community health, kinesiology, nursing, pharmacy, rheumatology, and surgery. We also have worked with researchers from other institutions, sometimes conducting CARDS meetings via teleconference. Nurses constitute the largest proportion of researcher teams (58%, $n = 23$) who have used the CARDS service and have included nursing faculty, doctoral students, and nurses working in community settings. Nurse researchers have brought a variety of materials and plans to the CARDS for their review and feedback, including recruitment materials (brochures, flyers, letters, scripts); study processes (recruitment plans, interview processes, website development); focus group protocols; interview and survey questions; study information sheets; and consent forms.

We sometimes have fielded inquiries from researchers who were interested in lay review of their materials but were skeptical that lay advisors who were not members of their particular target population could provide useful feedback. Materials used in human subjects research—recruitment notices, information sheets, questionnaires and other instruments, consent forms—share a common set of challenges for members of the public due to their technical language, dense presentation of information, embedded assumptions, counter-intuitive organization, and academic tone. As members of the general public with specific training on how to give feedback to researchers, the CARDS can offer a uniquely fresh perspective on research materials, no matter the specific content of the documents. The CARDS have provided useful feedback on materials for a wide range of health studies, despite having limited or no personal experience with many of the topics that researchers present. At one of our most memorable meetings, no women in the CARDS were able to attend, so five male CARDS provided feedback on a doctoral student's survey about dysmenorrhea. We introduced the survey by reminding the CARDS that we all have experience with pain and discomfort, and we asked them to think about the survey questions in the context of their personal pain experiences. Based on CARDS feedback, the student made substantial revisions to her survey.

Program Processes

Member orientation. One of the key features of the CARDS is the orientation program. Many CABs provide minimal orientation or training for board members (Albert Einstein College of Medicine, The Bronx Health Link, & Community-Campus Partnerships for Health, 2012). In contrast, the focus of the CARDS orientation program is hands-on practice with research materials. We designed an interactive group orientation to help the CARDS develop

Table 1. Responsibilities of Community-Academic Partners for CARDS® Program

Community Center Staff	WINRS Staff	CARDS®
Recruit people who use center services to participate in CARDS program	Design, deliver orientation program for new CARDS	Complete orientation to develop skills for giving feedback to researchers
Schedule community center facilities for meetings, orientations	Conduct outreach to identify guest researchers for CARDS meetings	NA
Arrange child care, transportation for monthly meetings; serve as contact person for members to confirm attendance	Meet with researchers to prepare agenda, materials for meetings; send agenda, meeting reminder to CARDS	RSVP for monthly meetings; read meeting announcement with description of research topic and materials
Participate in CARDS meetings	Facilitate CARDS meetings; take notes; disburse member stipends	Provide feedback to guest researchers at meetings
Invoice UW-Madison School of Nursing for facility fee	Write summary reports, revised materials for researchers; complete post-meeting evaluation surveys with guest researchers	NA
Problem-solve with members to address barriers to their participation	Work with liaisons, members to support participation and sustain program	As needed, meet privately with program staff to address issues or problems related to participation
Participate in program evaluation	Design and lead program evaluation	Participate in individual interviews and group discussions to evaluate program

Note. WINRS, Wisconsin Network for Research Support; UW-Madison, University of Wisconsin-Madison; NA, not applicable.

and practice the skills needed to participate effectively at meetings. Our orientation program emphasized content that is directly relevant to the work that board members will do and gives members experience reviewing and providing feedback on recruitment flyers and letters, consent forms, and focus group questions. Completion of the 2.5-hour orientation is a requirement for membership in the CARDS and provides meaningful preparation for respectful, productive interactions with researchers.

Preparation of guest researchers. Each CARDS group has met monthly at the community centers with guest researchers. Several weeks before the meeting, WINRS staff sent the scheduled researcher a timeline of steps to prepare for the meeting, along with a link to a short web-based survey requesting key information about the researcher's desired outcomes from the meeting. WINRS staff then engaged in a series of email messages and a telephone call or face-to-face meeting with the researcher to plan and prepare for the meeting.

During the planning meeting, WINRS staff and guest researchers reviewed answers to the web-based survey and discussed possible materials for the CARDS meeting. Researchers often brought their staff members and students to planning meetings and CARDS meetings. We select the materials that can be thoroughly discussed during a 90-minute CARDS meeting and develop a detailed working agenda. Without this preparation, researchers may have unrealistic expectations of how many documents can be reviewed during a meeting. During the planning meeting, WINRS staff shared advice on working with lay advisors. For example, we reminded researchers to use plain language, and we helped them prepare brief, straightforward explanations of their research interests. We also encouraged researchers to gracefully acknowledge all suggestions from the CARDS. Realistically, researchers may not want to or be able to follow certain suggestions, but thanking the CARDS for all suggestions helps CARDS feel that their voices are heard and welcomed. Decisions about how to use CARDS feedback can be made by researchers after they attend meetings, when they can decide whether to incorporate specific suggestions fully, partially, or not at all.

As part of meeting preparation, we discussed fees and payment arrangements with the guest researcher. We bundled our costs for WINRS and community center staff time, CARDS stipends, and other costs for the CARDS service into a fee of $1964, for a 90-minute meeting, preparatory work with the guest, and post-meeting summary documents. As required by the UW-Madison, our fee structure was based on a cost-recovery model; the fee for the CARDS service covered our costs for providing the service but did not generate profit. Researchers typically paid the fee with grant funds. For doctoral students or junior faculty without funding, we offered a reduced fee.

After we met with guest researchers, we sent a brief meeting announcement to the CARDS to provide information on the guest researchers, including their focus areas, materials they would present at the meeting, and type of help they would like from the CARDS. The announcement also included a personal statement from the guest researcher about why she or he cared about the research topic. We worked closely with researchers to help them craft personal statements. The CARDS have told us that learning about a researcher's personal motivations for doing research has helped them overcome their stereotypes of researchers as cold academics who use research participants to serve their own ends.

Meeting structure and practices. We have followed a structured sequence of activities at each meeting. We started each meeting with an opening question. Everyone at the meeting participated in a round-robin sharing of names and brief responses to the opening questions. Our opening questions gave everyone at the meeting a chance to share something about themselves. The activity helps to break stereotypes that researchers and lay community members may hold about each other. Over time, the sharing that occurs with the opening question helps group members establish personal connections and build a sense of community. We often crafted opening questions that were related to the research topic that we would discuss at the meeting. For example, our opening question for a meeting with a researcher studying nasal irrigation was "Please say your name, and tell us one favorite home remedy for dealing with a stuffy nose or other sinus problems."

After the opening question, guest researchers introduced themselves briefly, using plain language to explain the goal of their research and why it is important. We used the remaining 75 minutes of the 90-minute meeting to discuss the researcher's materials. The structure of the discussion was standardized and used three steps to elicit feedback from the CARDS:

1. The researcher describes how and where a prospective study participant would encounter the research materials, for example, mailed letter to a home address; flyer posted in a primary care clinic; consent form presented in a community setting; website that the participant will access from a home computer.
2. We ask guest researchers to read their materials aloud, several lines at a time, to facilitate full participation of everyone present, regardless of literacy level.
3. WINRS staff facilitate a section-by-section review of the materials. After a section is read, the CARDS offer comments. We take detailed notes and write CARDS feedback on a flip chart, a practice that helps to affirm the value of everyone's contribution to the discussion. After finishing review of one section, we move to the next section and repeat the process.

Post-meeting products and survey. During the first 2 years of CARDS meetings, we provided a summary report to researchers who attended meetings. The 1–3 page report summarized the overall feedback from the CARDS on the research materials reviewed at the meeting and highlighted specific issues related to the content, language, organization, and format of the materials. However, when researchers later shared their revised materials with us, we saw that they often did not translate CARDS feedback into concrete changes to their materials, although they rated the value of the meetings very highly. Consequently, we began to deliver revised versions of their project materials to all guest researchers, to make it easy for them to implement recommendations from the CARDS. Researchers consider and balance numerous factors when designing their study materials, and we encouraged researchers to use the CARDS-revised materials in whatever way makes the most sense for them, based on their experience and expertise. To complete our consultation, we sent a post-meeting online survey to all researchers who attend CARDS meetings to ask them to evaluate their experience.

Program Outcomes

CARDS® Insights and Recommendations

The value of the CARDS lies in their ability to provide researchers with fresh insights and feedback that academic or professional colleagues immersed in research may not be able to offer. As the concept of health literacy has permeated health sciences research, researchers have developed more awareness of how using technical jargon can undermine subject recruitment. Use of readability tools such as SMOG (National Cancer Institute, 1989) or the Flesch–Kincaid tool in word processing programs can be helpful for improving readability. Researchers also sometimes rely on colleagues, graduate students, high-level stakeholders, or IRBs to identify glaring problems in research materials. While these resources can be helpful, they may not be sufficient for ensuring that materials are acceptable and inviting to the general public. Over 5 years of monthly meetings, the CARDS have highlighted several key characteristics of research materials that may turn people away from participating in research.

Passive language. Plain language guidelines highlight the importance of using active language instead of passive language ("staff on my research team will ask you several questions" instead of "you will be asked several questions"). Passive sentences lack the clarity of active sentences because they do not clearly identify the actor. The CARDS highlighted other problems with passive language, describing it as confusing, impersonal, and evasive. For them, passive language in a research document raised questions and reservations about who is behind the research project, the true motives for the project, and what researchers will do with participants and their information. Passive language pervades consent forms, and it particularly troubled the CARDS when it appeared, as it does routinely, in material related to privacy and confidentiality. Vague assurances that "your information will be stored in a secure location" aroused their suspicions; they wanted to know exactly who would take responsibility for protecting their information.

Specialized use of everyday language. The CARDS provided feedback of a nuanced nature that highlights issues not measured by readability tools. The CARDS repeatedly pointed out things that were confusing, patronizing, or even offensive or frightening to potential research participants. For example, even though researchers may scrupulously avoid using technical jargon, they

often use everyday language in a highly specialized manner. The CARDS said that words such as "data," "procedures," "health outcomes," "technique," and "investigate" sounded ominous or threatening. Even the word "study," universally used by researchers in recruitment materials, can evoke images of being subjected to dangerous tests or used like a guinea pig. The CARDS strongly preferred "project" as a non-threatening alternative. When referring to research subjects, the CARDS strongly preferred the phrase "project participants," which sounds inviting, inclusive, and less scary than the off-putting, objectifying "subjects." Table 2 lists additional examples of how the CARDS interpreted common research language.

When we met with researchers to prepare for CARDS meetings, we explained that the CARDS may occasionally suggest alternative language that an IRB will reject. To our knowledge, these instances were infrequent and did not limit the value of the service. At the time of this writing, we were collaborating with an IRB workgroup that is charged with improving templates for informed consent. The working group has attended three CARDS meetings to learn the kinds of information about research that are important to the non-academic community and to help craft clearer language for consent documents. In the coming year, the working group expected to pilot new templates that incorporate CARDS feedback.

Perceived tone of documents. The CARDS noted several other key considerations for making documents accessible and inviting. The overall tone of a document can serve to engage or disengage potential participants. For example, although best practices in health education often recommend repeating key messages in documents, the CARDS said that redundant language feels insulting, by implying that they are not smart enough to understand something the first time that it appears in a document. In addition, materials that sound non-judgmental may be more likely to engage potential participants, especially for studies that address health behaviors. The CARDS often used the term "gentler" when recommending alternative language. For example, when researchers asked about tobacco use, the CARDS noted that the simple question "Do you want to quit smoking?" is a complex, sensitive question, and that asking a "yes" or "no" question may not be appropriate. The CARDS suggested, "Are you thinking about cutting back or quitting?" as a gentler alternative that raises the smoking issue without provoking defensiveness by implying that the person is doing something wrong.

Requests for demographic information. Demographic questions are standard items in many research instruments, and members of the public also frequently encounter them in the context of consumer research or participation in various programs. Despite the ubiquity of demographic questions, they may provoke negative reactions due to their highly personal nature. The CARDS told us repeatedly that they expected

Table 2. What Researchers Say, What CARDS® Hear, and What CARDS Recommend

What Researchers Say	What CARDS Hear	What CARDS Recommend
My colleague is the PI on this study.	I work with a private investigator who will poke into your personal business.	I work with Xxx Xxxx. (S)he is the lead researcher on this project.
Data will be collected about your lifestyle.	We will violate your privacy and make judgements about your personal life.	My research team will ask you some questions about a typical day for you.
Participants will take part in several procedures.	Participants will have scary, invasive medical acts performed on them.	Participants will be involved in several activities, including. . .
We will monitor your progress during this study.	We will track you with something like an electronic ankle bracelet during this study.	We will keep in touch with you during the project.
This study will test an experimental technique for treating sinus infections.	You will be a guinea pig for something dangerous, untried, and invasive.	This project will test a new way of treating sinus infections.
We would like to include your name in our study registry.	We want to put your name on a registry, like a sex offender registry!	We would like to add your name to a list of people who might like to be part of future projects.
Your input will contribute to the design of an intervention to support couples in parenting their babies.	You will help the research team confront couples who are having problems with parenting.	Your input will help us develop a new program to support parents as they care for their babies.
This study aims to improve how we provide care to people with type 2 diabetes.	Being in this study will improve your diabetes care.	We would like to hear about your experiences to help us figure out how to improve care for people with type 2 diabetes.
We will consider your individual home environment and search for assessable risks for falls.	We will judge you and your home and snoop into your personal belongings.	We will use a checklist to look for risks that you can change to prevent falls.
To manage your blood pressure, be more active and drink less alcohol.	We assume that you are not active and that you drink too much.	To manage your blood pressure, be active and limit your intake of alcohol.

researchers to use demographic information to stereotype people and present them in a bad light. The CARDS shared personal stories about skipping demographic questions, providing false answers, or declining to complete an instrument altogether if it includes demographic questions. They recommended that researchers ask only those questions that are critical for answering the study questions. In addition, the CARDS recommended that researchers provide a short, straightforward introduction to demographic questions to explain why the researcher needs the requested information. Explanations that simply invoke grant requirements for demographic information may be insufficient and potentially offensive; rather, a thoughtful explanation for asking demographic questions can explicitly acknowledge that demographic questions are personal and offer a straightforward rationale for asking them.

Lessons Learned on Engaging Hard-to-Reach Groups

When we initiated the CARDS program, we deliberately sought participants outside of the academic environment and not situated in positions of power. Our community center liaisons intentionally recruited people not typical of advisory boards. Many of them had had challenging life experiences, including poverty, homelessness, long-term underemployment, and chronic health problems. We structured the CARDS program to minimize barriers to participation and to demonstrate our respect and appreciation for all members.

Our use of conscious practices that engender trust and help people feel valued helped us to achieve outstanding retention of our lay advisors. Six of the current members (40%) have participated from 2010 to the time of this writing, and the remaining nine members had participated since 2012. Half of the former members who dropped out of the groups from 2010 to 2012 ($n = 6$) left because they moved out of the area, changed their work schedules, or experienced re-incarceration. Attendance has been very consistent. Since 2013, when we standardized many of our meeting processes, aggregated attendance for the program has been 81% (486 instances of member present for meeting/601 total opportunities to attend). Attendance for individual CARDS over this period has ranged from 58% of meetings to 97%.

Consistent staffing at both WINRS and the community centers has been a critical component of developing trusting relationships with our members and retaining them in the group over time. The same WINRS staff have facilitated every CARDS meeting from 2010 to the time of this writing, and the CARDS liaisons typically have stayed in their positions for at least 2 years. Consistent staffing helped us to develop trusting, long-term relationships with our members, which in turn supported honest dialogue and problem solving with members when challenges arise. On several occasions, we met with individual CARDS to address problems related to inappropriate behavior toward a program participant or insufficient participation in meeting discussions. The orientation that all CARDS complete served as a useful point of reference for constructive discussions about performance and expectations.

During our first few years, we often wrote short individualized notes to send to the CARDS after meetings, to thank them for their contributions. We called these notes "affirmations" because they affirmed something unique and positive that the person brings to the group. We also have held annual celebrations with the CARDS. Our celebrations have included meals, holiday treats, and small tokens such as gift cards.

For some of our members, applying for jobs is a frequent and difficult task. We have provided tangible support with letters of reference and assistance with resume writing. We have written letters of reference for members based on their work with the CARDS and provided resume templates that highlight the unique skills and tasks of the CARDS. With the advent of web-based systems for providing references, we have also completed on-line surveys to support job applications for some of the CARDS.

Program Evaluation by the CARDS®

In 2014, we completed a program evaluation with the CARDS. We conducted separate focus groups at each community center to explore members' opinions and attitudes in four areas: their orientation to the program; their work at CARDS meetings; factors that contributed to their retention in the program; and changes in how they viewed research and researchers. The CARDS valued the orientation and particularly emphasized the impact of group training activities on communicating respectfully and giving effective feedback. They also appreciated the orientation as a chance to experience what they would do at meetings, and several members credited the orientation with giving them confidence that they could do the job. The CARDS described multiple characteristics of the monthly meetings that make the work meaningful and enjoyable. These included learning about research topics that were relevant to them and their community and having a chance to give advice directly to researchers.

The most prominent theme throughout the focus groups was the sense of community that motivated the CARDS to stay in the program. They used words such as respect, good will, support, camaraderie, and fellowship to explain why they stayed in the program. They also described how our standard meeting practices such as the opening question, use of the flip chart, meeting snacks, and non-hierarchical facilitation contributed to a sense of community. Another major factor related to retention was the sense of service to others. As one

member said, "I like being able to help others. We may be helping folks who will never even know we helped them." Finally, the CARDS discussed how their attitudes toward research and researchers have shifted. They said that the opportunity to work regularly with researchers had helped them respect researchers, rather than mistrust their motives. Some CARDS also indicated that they were more interested in participating in research themselves: "Now I feel it's important to participate in research. Researchers are doing it for the community; it's bigger than just one person."

Researchers' Evaluation of CARDS® Service

Since 2012, all guests who attended CARDS meetings received an email 1 week after the meeting inviting them to complete a brief web-based evaluation survey. To minimize the response burden on researchers, the survey included just a few key questions to assess guests' satisfaction with the overall CARDS service, including pre-meeting preparation, and post-meeting reports. Guests at CARDS meetings rated the service very highly. Ninety-seven percent ($n = 70$) of survey respondents indicated that they felt sufficiently prepared by WINRS to meet with the CARDS. Guests typically brought one or two items for review, and they most frequently brought data collection materials such as survey or focus group questions (51% of researchers, $n = 37$) and recruitment materials (37%, $n = 27$). A large majority of guests (90%, $n = 65$) planned to change their materials based on CARDS feedback. (Several respondents self-identified as project staff that did not have authority to make changes to research materials.) Ninety-nine percent of survey respondents ($n = 71$; 1 missing response) indicated that they would recommend the service to colleagues. Table 3 displays findings related to guests' perceptions of the utility of the CARDS program.

When we initiated the program, we wondered whether regular contact with researchers would acclimate the CARDS to research language and over time diminish their value to researchers. To preserve the lay perspective of the CARDS, we have maintained a schedule of meeting only once monthly at each center. Researchers' evaluations of the CARDS service have been favorable across the lifespan of the program, suggesting that the CARDS have continued to offer valuable feedback. Findings related to tests of a target population's response to the CARDS' recommended changes in research materials are published elsewhere (Bowers, Jacobson, & Krupp, 2016).

Table 3. Guests' Perception of Utility of CARDS® Services, 2012–2016 ($N = 72$)

Which Services of the CARDS Program Were Particularly Useful?	*n*	%
Completing web-based survey to initiate planning	28	39
Meeting with program staff[a]	44	61
Presenting ideas and materials to CARDS	56	78
Getting feedback from CARDS	70	97
Receiving summary of CARDS feedback	65	90
Receiving revised study materials	60	83

Note. Percentages do not sum to 100% because respondents could select multiple options.
[a]New question added to survey in 2103.

Conclusion

Health sciences research should serve everyone. When researchers have study participants who reflect the rich diversity in our communities, our society has a real chance to develop policies and practices to eliminate health disparities. But current research suggests that equitable representation of diverse groups in research has yet to be achieved (Bonevski et al., 2014). Social justice is a central concern of the nursing profession, and nursing research literature includes numerous examples of studies using approaches such as CBPR to engage disenfranchised or hard-to-reach stakeholders in the research process (e.g., Perry & Hoffman, 2010; Stacciarini et al., 2011). In addition, many nurse researchers have designed or implemented interventions with input from community health workers or promotores (e.g., Nies, Troutman-Jordan, Branche, Moore-Harrison, & Hohensee, 2013; Whittemore, Rosenberg, Gilmore, Withey, & Breault, 2014).

The CARDS represent a new, unique model for engagement of people who are not prominent community representatives in the design of research. The model offers distinct advantages over other forms of public engagement. Although lay stakeholder engagement can take diverse forms, sometimes it can seem as if the options are all or nothing—either researchers conduct CBPR or they do nothing to seek community input. The CARDS model makes reaching hard-to-reach stakeholders feasible and convenient for researchers. The CARDS bring hidden voices into the research enterprise and make these voices easily accessible to researchers. CARDS is an innovative model for nurse researchers to support sustainable, meaningful engagement of hard-to-reach populations in research planning and activities; increase the diversity of groups represented in research studies; and ultimately help to reduce health disparities.

References

Albert Einstein College of Medicine, The Bronx Health Link, & Community-Campus Partnerships for Health. (2012). *Community IRBs & research review boards: Shaping the future of community-engaged research*. Retrieved from http://depts.washington.edu/ccph/pdf_files/Shaping_the_Future_of_CEnR.pdf

Bonevski, B., Randell, M., Paul, C., Chapman, K., Twyman, L., Bryant, J., ... Hughes, C. (2014). Reaching the hard-to-reach: A systematic review of strategies for improving health and medical research with socially disadvantaged groups. *BMC Medical Research Methodology*, *14*, 42. doi: 10.1186/1471-2288-14-42

Bowers, B. J., Jacobson, N., & Krupp, A. (2016). Can lay community advisors improve recruitment of underrepresented populations? *Research in Nursing & Health* [advance online publication]. doi: 10.1002/nur21752.

D'Alonzo, K. T. (2010). Getting started in CBPR: Lessons in building community partnerships for new researchers. *Nursing Inquiry*, *17*, 282–288. doi: 10.1111/j.1440-1800.2010.00510.x

Fernandez-Pena, J. R., Moore, L., Goldstein, E., Decarlo, P., Grinstead, O., Hunt, C., ... Wilson, H. (2008). Making sure research is used: Community-generated recommendations for disseminating research. *Progress in Community Health Partnerships*, *2*, 171–176. doi: 10.1353/cpr.0.0013

Ford, J. G., Howerton, M. W., Lai, G. Y., Gary, T. L., Bolen, S., Gibbons, M. C., ... Bass, E. B. (2008). Barriers to recruiting underrepresented populations to cancer clinical trials: A systematic review. *Cancer*, *112*, 228–242. doi: 10.1002/cncr.23157

GreenMills, L. L., Davison, K. K., Gordon, K. E., Kaigang, L., & Jurkowski, J. M. (2013). Evaluation of a childhood obesity awareness campaign targeting Head Start families: Designed by parents for parents. *Journal of Health Care for the Poor & Underserved*, *24* (2 Suppl), 25–33. doi: 10.1353/hpu.2013.0096

Hasnain-Wynia, R., & Beal, A. C. (2014). Role of the patient-centered outcomes research institute in addressing disparities and engaging patients in clinical research. *Clinical Therapeutics*, *36*, 619–623. doi: 10.1016/j.clinthera.2014.04.005

Institute of Medicine. (2011). *Public engagement and clinical trials: New models and disruptive technologies: Workshop summary*. Washington, DC: National Academies Press.

Israel, B. A., Parker, E. A., Rowe, Z., Salvatore, A., Minkler, M., Lopez, J., ... Halstead, S. (2005). Community-based participatory research: Lessons learned from the centers for children's environmental health and disease prevention research. *Environmental Health Perspectives*, *113*, 1463–1471.

James, S., Arniella, G., Bickell, N. A., Walker, W., Robinson, V., Taylor, B., & Horowitz, C. R. (2011). Community action boards: An innovative model for effective community-academic research partnerships. *Progress in Community Health Partnerships*, *5*, 399–404.

Joosten, Y. A., Israel, T. L., Williams, N. A., Boone, L. R., Schlundt, D. G., Mouton, C. P., ... Wilkins, C. H. (2015). Community engagement studios: A structured approach to obtaining meaningful input from stakeholders to inform research. *Academic Medicine*, *90*, 1646–1650. doi: 10.1097/acm.0000000000000794

Kitterman, D. R., Cheng, S. K., Dilts, D. M., & Orwoll, E. S. (2011). The prevalence and economic impact of low-enrolling clinical studies at an academic medical center. *Academic Medicine*, *86*, 1360–1366. doi: 10.1097/ACM.0b013e3182306440

National Cancer Institute. (1989). *Making health communication programs work*. Bethesda, MD: National Institutes of Health, US Department of Health and Human Services.

National Institute on Minority Health and Health Disparities. (2016). *About NIMHD: Vision and mission*. Retrieved from http://www.nimhd.nih.gov/about/visionMission.html

Newman, S. D., Andrews, J. O., Magwood, G. S., Jenkins, C., Cox, M. J., & Williamson, D. C. (2011). Community advisory boards in community-based participatory research: A synthesis of best processes. *Preventing Chronic Disease: Public Health Research, Practice, and Policy*, *8*(3), A70.

Nies, M. A., Troutman-Jordan, M., Branche, D., Moore-Harrison, T., & Hohensee, C. (2013). Physical activity preferences for low-income sedentary urban African American older adults. *Journal of Gerontological Nursing*, *39*, 20–29. doi: 10.3928/00989134-20130408-01

Paskett, E. D., Reeves, K. W., McLaughlin, J. M., Katz, M. L., McAlearney, A. S., Ruffin, M. T., ... Gehlert, S. (2008). Recruitment of minority and underserved populations in the United States: The centers for population health and health disparities experience. *Contemporary Clinical Trials*, *29*, 847–861. doi: 10.1016/j.cct.2008.07.006

Perry, C., & Hoffman, B. (2010). Assessing tribal youth physical activity and programming using a community-based participatory research approach. *Public Health Nursing*, *27*, 104–114. doi: 10.1111/j.1525-1446.2010.00833.x

Pinto, R. M., Spector, A., Rahman, R., & Gastolomendo, J. D. (2013). Research advisory board members' contributions and expectations in the USA. *Health Promotion International*, *30*, 328–338. doi: 10.1093/heapro/dat042

Schroen, A. T., Petroni, G. R., Wang, H., Gray, R., Wang, X. F., Cronin, W., ... Slingluff, C. L., Jr. (2010). Preliminary evaluation of factors associated with premature trial closure and feasibility of accrual benchmarks in phase iii oncology trials. *Clinical Trials*, *7*, 312–321. doi: 10.1177/1740774510374973

Silvestre, A. J., Quinn, S. J., & Rinaldo, C. R. (2010). A twenty-two-year-old community advisory board: Health research as an opportunity for social change. *Journal of Community Practice*, *18*, 58–75. doi: 10.1080/10705421003766685

Stacciarini, J.-M. R., Wiens, B., Coady, M., Schwait, A. B., Péérez, A., Locke, B., ... Bernardi, K. (2011). CBPR: Building partnerships with Latinos in a rural area for a wellness approach to mental health. *Issues in Mental Health Nursing*, *32*, 486–492. doi: 10.3109/01612840.2011.576326

Stewart, M. K., Felix, H. C., Olson, M., Cottoms, N., Bachelder, A., Smith, J., ... Greene, P. G. (2015). Community engagement in health-related research: A case study of a community-linked research infrastructure, Jefferson County, Arkansas, 2011–2013. *Preventing Chronic Disease: Public Health Research, Practice, and Policy*, *12*, 140564. doi: 10.5888/pcd12.140564

Strauss, R. P., Sengupta, S., Quinn, S. C., Goeppinger, J., Spaulding, C., Kegeles, S. M., & Millett, G. (2001). The role of community advisory boards: Involving communities in the informed consent process. *American Journal of Public Health*, *91*, 1938–1943.

US Department of Health and Human Services. (2016). *About Healthy People*. Retrieved from http://www.healthypeople.gov/2020/about/default.aspx

Whittemore, R., Rosenberg, A., Gilmore, L., Withey, M., & Breault, A. (2014). Implementation of a diabetes prevention program in public housing communities. *Public Health Nursing*, *31*, 317–326. doi: 10.1111/phn.12093

Acknowledgments

The Wisconsin Network for Research Support (WINRS) and the CARDS® are funded by the University of Wisconsin-Madison School of Nursing and the Clinical and Translational Science Award (CTSA) program, through the NIH National Center for Advancing Translational Sciences (NCATS), grant UL1TR000427. The content is solely the responsibility of the authors and does not necessarily represent the official views of the NIH. The authors thank current and former CARDS members and staff at the Lussier Community Education Center and Goodman Community Center for their contributions to the CARDS program.

The Power of The Personal: Breaking Down Stereotypes and Building Human Connections

Gay R. Thomas, Betty L. Kaiser, and Kaitlin Svabek

Narrative Inquiry in Bioethics, Volume 7, Number 1, Spring 2017, pp. 27-30 (Article)

Published by Johns Hopkins University Press
DOI: https://doi.org/10.1353/nib.2017.0010

On a weekday night, every month, twelve people meet around a table at a community center in Madison, Wisconsin. The group includes people who are homeless, previously–incarcerated, unemployed, handling chronic mental or physical challenges and several health science researchers. Someone walking by the room might notice the intense energy, lively debate, engaged participation, reams of flip chart paper, and wonder, "What's going on??"

This has been our reality every month for over six years. The people *giving* advice are the Community Advisors on Research Design and Strategies (CARDS)®, community members from diverse racial, socioeconomic, and educational backgrounds. The people *getting* advice are researchers who want candid feedback about how to make their materials more engaging, easier to understand and more actionable from people often labelled "hard–to–reach." And the people planning the meetings are our staff with the Wisconsin Network for Research Support (WINRS), a patient and community engagement resource.

We started the CARDS® in 2010 as a partnership between the University of Wisconsin–Madison School of Nursing, Lussier Community Education Center, and Goodman Community Center. With initial funding from a 3–year National Institutes of Health grant, our project was a response to the painful reality of persistent health disparities in our country and to the fact that health sciences research has not successfully engaged the full breadth of our country's wonderful diversity. What's the connection, we wondered? How can research appeal to a broader group of people? If all Americans did participate equally in health sciences research, would this move the needle on health inequality? With our community partners, we deliberately recruited CARDS® from groups of people most affected by health disparities and least represented in research projects. We provide an interactive orientation for all CARDS® and pay members for each meeting they attend. We also compensate the community centers for staff time and meeting space.

In the process of bringing unheard voices into the research enterprise, we have learned a lot about how to revise research activities and materials to engage a broader audience. We've also learned some interesting things about the stereotypes and assumptions that researchers make about "hard to reach" people, the assumptions that lay people make about researchers, and how to overcome prejudices that make it hard to connect with each other.

What are some common researcher assumptions? "I've spent my whole professional life researching this disease, what can I learn from people who don't really know anything about this topic?" "I know what I will get—people telling me to 'dumb down' my materials." "The feedback I

Access provided by Arizona State University (19 Jul 2017 21:04 GMT)

get from colleagues is sufficient." And community members have their own pre–conceptions: "I think research projects are a scam. Researchers have a hidden agenda and just are trying to help themselves." "Lots of researchers are white—they don't want to hear my ideas." "Research? Nope, I don't want to be a guinea pig!" "Most researchers are uppity and just talk down to us." And these are just the comments conveyed to our WINRS team directly. Obviously, other prejudices and stereotypes also divide us—based on how we dress, talk, our skin color, the condition of our teeth, etc.

Imagine trying to communicate in a room where these two worlds collide. On one side: I see you as less educated, less smart, and not able to offer me anything useful. On the other: I see you as sneaky, self–interested, arrogant and out to take advantage of me. How do we find common ground? How can researchers get the advice they need to make their projects more accessible, appropriate and engaging? How can we create a space where lay people feel free to share important feedback?

Two specific practices we use to break down divisive assumptions and stereotypes demonstrate the power of "The Personal." The first practice is to start each CARDS® meeting with an opening question that everyone at the meeting answers. This may seem straightforward, but learning what makes an effective opening question has been an experience of considerable trial and error.

At one meeting, the guest was a young researcher who appeared very stiff and ill at ease at the outset. Our opening question was: "Think about a time when you took care of someone else—a child, relative or friend. Tell us what made you good at taking care of that person?" As we went around the room, we heard from a woman caring for her grandma, who described the patience (and humor!) required to deal with increasing forgetfulness. We heard about stepping up to be on call 24/7 for an ailing friend and making the decision to move in with aging parents. When it was the researcher's turn, she started by simply holding up her hands. People shifted uncomfortably in their seats, wondering if she was giving up on the meeting before it had even started. Then she spoke: "I have 3 young children," she said gently. "I've always felt like human touch is one of the most caring gifts we can give each other. Every time I bathe my kids, wipe their noses or their tears, wash a scrape . . . touching them in a tender, loving way seems like one thing that makes me good at taking care of them." By the time she finished this very short statement, there were plenty of damp eyes and warm smiles around the table, and we were all looking at each other with a very different perspective. The power of "The Personal."

At another meeting, we asked, "Looking back on your childhood, what is one *good* memory that really sticks with you?" One of our CARDS® has struggled with drug addiction and shows many of the physical ravages of this difficult history. It would be hard for most people to look at this person and not jump to conclusions based on outward appearance. When it was his turn to answer the opening question, he said: "I grew up in New York. I had the best granny in the world. Every year, she got me all dressed up and took me to see the Christmas show with the Radio City Rockettes. Every single year! She said I would never miss a Rockettes Christmas show—and I never did." The impact on the researcher (and the rest of us) was amazing. To see him glow with this happy memory and to unexpectedly glimpse the excited, dressed–up little boy still inside this grown man was transformative. The power of "The Personal."

But not just any opening question works! Here are some lessons we've learned.

Some opening questions are simply boring, and do not help us connect with each other. When we had dementia researchers at a CARDS® meeting, we said, "Tonight we are going to talk about research on memory. What is one trick you have for remembering or keeping track of things?" Not surprisingly, the answers were all basically identical: "I write lists." "I have a special place where I put important things, like my keys." No one shared anything uniquely personal—so no personal connections were established and no stereotypes were really challenged.

We've also learned that while "The Personal" is powerful, it needs to be used thoughtfully. Some opening questions can unintentionally derail the

group with negative energy, making it harder to get on a productive track. One night we said, "We are going to talk about home care after surgery. That got us thinking about scars—we all have scars from accidents, injuries or wounds. Please tell us a story about one scar you have." Although we prepared a staff member to model a light-hearted memory of a scar she got while playing with her beloved baby sister, the following stories took a darker turn. We heard about a stillborn baby, injuries from the Vietnam War and serious work–related accidents. Soon the group was extremely somber, with a palpable sadness around the table created by stories that led us into our own separate worlds of grief.

Lesson learned—craft a thoughtful opening question likely to draw out stories and experiences that engender positive emotions and reinforce human connections. This not only helps us to see past the stereotypes and assumptions that divide us, it also reduces our own barriers of discomfort, nervousness and feeling out of place. It's hard to connect with a stereotype. It's hard *not* to connect with a good personal story. As one of our CARDS® told us, "I love the opening questions. They help us accept the researcher. When we share life experiences, we see each other as human."

The second personal practice that breaks down stereotypes is using five minutes of meeting time for guest researchers to explain their personal motivation for their research. The CARDS® have repeatedly told us how critical it is to understand why a researcher is focused on a specific topic: "Are they just doing this for the money or do they actually care about this problem?" The fundamental truth we've learned is "people don't care how much you know until they know how much you care." However, when we first started asking researchers to comment on what motivates them to do their research, they generally discussed "gaps in the literature" or the "iterative nature" of the research process. Researchers are trained to be objective and scientific—not to talk from the heart when discussing their program of research. But we have found that with some individualized coaching, researchers of all kinds can benefit from the power of "The Personal."

Now we specifically ask researchers to reflect on why they are passionate about their research and to answer this question in a short survey before coming to a CARDS® meeting. We follow up with each researcher and together prepare a short "script" for the researcher to share at the meeting—expressing in 2–3 sentences why they have a personal stake in their research topic. Consider the difference between "I'm part of a multidisciplinary team facilitating innovative science, targeting the prevention and treatment of this disease with novel research projects . . ." versus "This research is important to me because of my own family history with this disease. I am personally committed to making a difference in the prevention and treatment of this disease." OR "I focus on this research because scientists still don't understand the most cost–effective way to treat this condition . . ." versus "I am a physician who treats people with this condition. I have seen my patients struggle with both the terrible symptoms of this disease and with the high costs to treat it. I hope what we learn in this project will help my patients and the many others whose lives are damaged by this condition."

Some of the most poignant comments we have heard in evaluations with the CARDS® are about how their attitudes towards researchers have shifted as a result of the power of "The Personal." As one of the CARDS® put it, "I get really moved when researchers tell us why they study what they do—the stories of grandparents, other family members, patients and so on. You can see that they genuinely want to make the world a better place." Another said, "I used to think that researchers were 'off in another universe,' but now I see them as decent, caring human beings who are trying to do good things."

Researchers also have shared touching comments about being able to connect with these "hard to reach" people: "The single best thing about meeting with the CARDS® was getting feedback from community members we normally wouldn't have the opportunity to talk with." "Getting advice from people with 'lived experience' is more critical than we realized. Many researchers don't consider the perspective of participants. I really didn't

think about this issue seriously, but this meeting improved my materials *and* changed my attitude!"

These two practices, thoughtfully and consistently followed, enable us to harness the power of "The Personal"—breaking down barriers and building human connections that empower all stakeholders to participate in reducing health disparities and improving health outcomes for everyone.

Acknowledgements. The authors thank current and former CARDS® and staff at the Lussier Community Education Center and Goodman Community Center for their contributions to the CARDS® program.

Funding. The Wisconsin Network for Research Support (WINRS) and the Community Advisors on Research Design and Strategies (CARDS)® are partially funded by the University of Wisconsin – Madison School of Nursing and the Clinical and Translational Science Award (CTSA) program, through the NIH National Center for Advancing Translational Sciences (NCATS), grant UL1TR000427. The content is solely the responsibility of the authors and does not necessarily represent the official views of NIH.

For additional information about this article

https://muse.jhu.edu/article/664716

ADDITIONAL HOMEWORK FOR SESSION 2

LINK TO VIDEOS:

In addition to reading the preceding case study and the "the power of the personal" article, go ***HERE*** to watch these brief videos:

1. Five researchers share examples of how the CARDS(R) improved study recruitment and research outcomes *(7:14 min)*
2. Community Advisors at the Goodman Community Center (Madison, WI) share insights about research materials *(5:38 min)*
3. Community Advisors discuss personal benefits of participating in the CARDS(R) program *(7:25 min)*
4. Community Advisors at Lussier Community Education Center (Madison, WI) reflect on impact of being a CARDS(R) advisor *(7:13 min)*

Sample Agenda

90-minute Stakeholder Meeting

- **Location**
- **Date**
- **Time**

Item	Lead	Time	Purpose
Welcome/Announcements		**2 mins**	• Welcome • Good time to turn off cell phone • Review meeting tips • Other announcements as necessary
"Close the Loop"		**3 mins**	• Show impact/value of stakeholder input • Demonstrate transparency/ honesty in how input is being used
Opening Question/Intros		**10-15 mins**	• Get everyone talking/engaged • Use a question related to content of meeting, if possible • Focus on assets of all participants
Research Overview		**5 mins**	• Provide brief introduction to researcher/research topic • Offer insight into researcher's personal interest in topic • Help stakeholders personally connect with researcher

Group discussion			

"Set the Stage"		**5 mins**	• Help focus stakeholders to give relevant feedback
			• Provide key info, such as:
			  ○ Who is the audience? Who will see the material/ document (age, etc)?
			  ○ Where will they be when they see it? (at home alone, in an appointment with a health care provider)
			  ○ What does the research team want the target audience to DO as a result? What is your goal for this material?
			  ○ Any other key information?

Review Materials		**60-70 mins**	• Go through the materials section by section, reading each section aloud and asking for feedback.
			• Ask questions such as:
			  ○ What are your first impressions?
			  ○ Does this make sense?
			  ○ Is it clear, easy to understand?
			  ○ It is engaging/interesting?
			  ○ Is any important information missing?
			  ○ Is anything confusing, off-putting, scary or offensive?
			  ○ How can this material be improved?

NOTES FROM SESSION 2

Stakeholder Engagement Basics for Health Science Researchers:
Putting the Pieces Together

SESSION 3

SESSION 3, May 10

PRE-SESSION HOMEWORK:

1. Download and review the "HARPS Roadmap," ***How To Recruit and Retain Patient Stakeholders*** (HARPS); available ***HERE*** *(free registration will be required)*
2. Go through the roadmap, and click on the video prompts to watch videos associated with each section. To watch ALL of the videos, plan for about 80 minutes. If prioritizing, try to watch at least the following videos, which should take about 45 minutes:
   a. Who do I want as stakeholders? (section 1)
   b. What do I want my stakeholders to do? (section 2)
   c. How will I recruit patient stakeholders? (section 4)
   d. How will I prepare stakeholders for their work? (section 6)
   e. How can I facilitate effective meetings with my stakeholders? (section 8)
   f. How do I budget for stakeholder engagement? (section 10)

DURING AND AFTER SESSION:

1. Participate in CARDS® meeting
2. Complete personal reflections/reactions to CARDS® meeting experience *(we will disseminate a link to an online survey platform after the meeting)*

NOTES FROM SESSION 3

Stakeholder Engagement Basics for Health Science Researchers:
Putting the Pieces Together

SESSION 4

SESSION 4, June 14

PRE-SESSION HOMEWORK:

1. Download and review the ***Patient Advisor Toolkit 1: Orientation for Patient Advisory Committees*** (PAT-1); available ***HERE*** *(free registration will be required)*
2. Review the ***Patient-Centered Outcomes Research Institute (PCORI) website***; available ***HERE***. Of particular interest under "About PCORI" are PCORI's vision and mission statement as well as its new strategic plan.
3. Read the ***Strengthening Diversity in Research Partnerships: Knowledge to Action Guide*** by the Institute for Patient and-Family-Centered Care©; available ***HERE***
4. Read Largent et al, ***"Patient-engaged research: choosing the 'right' patients to avoid pitfalls"*** in the *Hastings Center Report* 2018, pages 41-49 in this booklet

Patient-Engaged Research

Choosing the "Right" Patients to Avoid Pitfalls

BY EMILY A. LARGENT, HOLLY FERNANDEZ LYNCH, AND MATTHEW S. MCCOY

Including patients' perspectives in the design and conduct of clinical research is thought to be useful for aligning health care with patients' needs and priorities. But which patients are engaged—and which should be? Engaging the "wrong" patients might even be detrimental.

The U.S. Congress's 2010 authorization of the Patient-Centered Outcomes Research Institute[1] represented an inflection point in the long normative push toward including patient perspectives in the design and conduct of clinical research: a major research sponsor would now require such patient engagement as a foundational condition of funding. Thus, PCORI is a useful case study for understanding and evaluating patient-engagement efforts more broadly. PCORI's mission is to "help . . . people make informed healthcare decisions, and improve . . . healthcare delivery and outcomes, by producing and promoting high-integrity, evidence-based information that comes from research guided by patients, caregivers, and the broader healthcare community."[2] One of PCORI's animating beliefs is that patients have knowledge and insights that researchers lack. Thus, research meaningfully informed by the patient perspective will be more relevant to the complex choices individuals face when it comes to their health care, allowing them to make better decisions in line with their own goals and priorities.

To ensure that the information resulting from PCORI-funded research is relevant to patients, PCORI eschews the "traditional health research" paradigm, in which investigators drive all aspects of research, in favor of one in which patients assume the role of research partner.[3] To effectuate this paradigm shift, PCORI requires that the research proposals it funds include patients and other stakeholders at every step of the research process—"from proposal development to research design and dissemination of the study results."[4] Patient engagement can take many forms, from offering information, advice, and

Emily A. Largent, Holly Fernandez Lynch, and Matthew S. McCoy, "Patient-Engaged Research: Choosing the 'Right' Patients to Avoid Pitfalls," *Hastings Center Report* 48, no. 5 (2018): 26-34. DOI: 10.1002/hast.898

feedback to serving as a coinvestigator participating in recruitment, data collection, and data analysis.[5]

Further empirical analysis is needed to determine the effects of patient engagement on research and whether PCORI's stated goals are being achieved.[6] Yet if we accept the premise that patient engagement can offer fresh perspectives that shape research in valuable ways, then at least two important sets of questions present themselves. First, *how* are patients being engaged—and how should they be engaged? PCORI has devoted substantial energy to addressing these questions, and it requires all applications to include an "engagement plan" detailing how patients will be engaged at each phase of research.[7] In addition, there is a small but growing body of research that examines different methods of patient engagement employed by PCORI-funded researchers.[8]

The second set of questions, which has received relatively less attention, is *which* patients are being engaged—and which patients should be? This neglect is somewhat surprising, given that the "who" question is conceptually prior to the question of "how." For if the "wrong" patients are engaged in research—even if they are engaged in the "right" ways—their input will have less value and might even be detrimental. In light of that possibility, we suggest that not all patient engagement should be treated as equal and, perhaps controversially, that patient engagement is not necessarily an unmitigated good.

Given these concerns, this article focuses attention on the "who" of patient engagement in research. First, we provide background on the rationale for patient engagement, underscoring the importance of ensuring the representativeness of engaged patients. Second, we present what little is known about patients engaged in PCORI-funded research. Third, we identify and discuss the ethical implications of ways in which current practices of patient identification and recruitment may lead to a lack of representativeness. These practices include reliance on the well-connected and well-informed, reliance on patients who are not well trained or well-informed, and reliance on patient advocacy organizations. Finally, we consider several strategies for addressing these pitfalls in order to maximize the positive goals of patient engagement. Patient engagement is intended to address the inability of researchers, funders, and others to fully represent patient views and priorities, but without sufficient attention, the patients selected for this role may still leave important gaps.

Goals and Motivations of Patient Engagement

Patient engagement is central to PCORI's mission,[9] but it is not, of course, an end unto itself. Rather, patient engagement is a means of ensuring that research is "patient centered, useful, and trustworthy," leading to "greater use and uptake of research results by the patient and broader healthcare community."[10] Why believe that patient engagement will have these salutary effects? The premise behind this idea is that patients are meaningfully different from researchers by virtue of their firsthand experience with a particular disease or condition, which in turn gives them unique knowledge and insights. For example, patients can help researchers better understand not only what it is like to live with a particular condition but what sorts of interventions might make the greatest contribution to their well-being. They can identify unmet needs facing patients as well as problems that cause existing interventions to fall short. Moreover, as relative "outsiders" to the research enterprise, patients can raise relevant questions and concerns about aspects of research that "insiders" on the research team might take for granted, such as what might make research participation more or less attractive for members of a particular patient community.

If the purpose of patient engagement is to help researchers better understand the perspectives of a given patient population, then the patients engaged should reflect the range of characteristics, experiences, and interests of the people in that population. We can think of representativeness in this context not as a statistical benchmark to be achieved but as a regulative ideal that is realized when a variety of voices are heard. The closer engaged patients come to reflecting the full diversity of a population of interest, the greater the likely value of their input. By contrast, when the engaged patients are a narrow and relatively homogenous sample of the people in a patient population, then their input will be of limited value and may even compromise research. We will examine the problems associated with a lack of adequate representation in patient engagement in greater detail below, but first we review what is known about who is presently being engaged in patient-centered outcomes research.

Patients are individuals with distinct traits, experiences, interests, goals, and relationships. A choice to engage some patients instead of others will have consequences for which perspectives inform research.

Who Is Being Engaged Now

Unfortunately, little is known about those currently engaged in patient-centered outcomes research. In 2017, Lauren Ellis and Nancy

Kass published findings from interviews of thirty-three patients engaged in PCORI-funded research, offering some of the most comprehensive demographic information available to date.[11] They found that "[m]ore than two-thirds of patients . . . were female; the majority were white, with a wide age range represented . . . . More than two-thirds were employed and held a college or graduate degree . . . . [M]ore than one-third had previously engaged in research."[12] This data has obvious limitations, coming as it does from a small qualitative study, but it suggests high levels of homogeneity among engaged patients across important parameters.

Slightly more is known about how patients are selected for engagement. Research suggests that many patients are recruited based on longstanding relationships with the principal investigator or through the PI's personal network. Ellis and Kass, for instance, found that in more than half of the projects they studied, the PIs and patients had "preexisting relationships, often from prior engagement or professional interactions, including working at the same institution or shared membership in an organization."[13] In other words, there is strong reliance on convenience sampling as the method to identify patients for engagement in research. Other investigators doing patient-centered outcomes research have reported forming new relationships for purposes of patient engagement—recruiting patients from clinics, national patient associations, local disease groups, their hospital's patient advisory council, or their professional network.[14] While some of these "new" connections seem to reflect outreach to individuals outside the investigators' existing circles, others do not. This data, too, is suggestive of relative homogeneity in the patients selected for engagement.

Potential Pitfalls

As the push intensifies to think of patients as coequals in the research process, we must think about the "who" of patient engagement in a more systematic and critical way. Patients are not a monolithic group. They are individuals with distinct traits, experiences, interests, goals, and relationships. Accordingly, a choice to engage some patients instead of others will have important consequences for which perspectives inform research. These choices not only affect the instrumental value of patient engagement but also have downstream ethical implications that deserve greater attention than they have received to date.

If the patients engaged do not adequately reflect the range of diversity within a target patient population, some perspectives may be emphasized while others get ignored. An apt analogy can be drawn to clinical trial recruitment: researchers seek to avoid collecting data from too homogenous a group because that will prevent them from generalizing the results in the desired way. Similarly, in patient-engaged research, if only a narrow range of patient voices are heard, then researchers will gain a partial—and potentially misleading—sense of the concerns and needs of patients in the population of interest. More problematically, this kind of skewed representation will often benefit the relatively well-off at the expense of socially marginalized groups, therefore "perpetuating disadvantage to other groups of patients whose perspectives continue to be excluded," as Ellis and Kass note.[15]

If research is guided by patient engagement, then which patients are involved will determine which questions are asked, how they are asked, and how data is interpreted to answer them. These decisions at the research phase can have far-reaching effects when research is used to shape health policy. For example, one of PCORI's national priorities is to address health disparities by "identifying potential differences in prevention, diagnosis, or treatment effectiveness, or preferred clinical outcomes across patient populations and the healthcare required to achieve best outcomes in each population."[16] Yet if patient-centered outcomes research is disproportionately influenced by the perspectives of a narrow, relatively well-off segment of patients, then the treatment and policy decisions it informs are likely to reflect this bias and may fail to address or even exacerbate existing disparities. Ultimately, then, concerns about the representativeness of engaged patients are closely associated with concerns about justice and who benefits from research.

There are at least three ways in which practices of patient identification and recruitment currently used by PCORI-funded researchers can lead to lack of representativeness.

Relying on patients who are well-informed and well connected. As detailed above, engaged patients are often recruited via convenience sampling. Patient engagement requires commitment from both the researchers and the engaged patients to maintain contact and participation, and trust has been described as a key factor in sustaining engagement over time.[17] Thus, it is not surprising that PIs would turn to individuals with whom they have preexisting relationships or who are referred to them via their professional networks. Longstanding relationships are likely to be based on some degree of trust and to allow PIs insight into the potential quality of a working relationship. Furthermore, researchers may find it difficult to establish new partnerships with patients under the time pressure of writing a research proposal.[18] Similar considerations explain why investigators often collaborate with professional colleagues they already know rather than initiating new collaborative relationships.

Yet patients engaged via convenience sampling are unlikely to be sufficiently representative of the range of patient experiences relevant to the research. For instance, we might reasonably hypothesize that convenience sampling yields engaged patients with higher health literacy than the general population.

Health literacy is, according to a report from the Institute of Medicine, "[t]he degree to which individuals have the capacity to obtain, process, and understand basic health information and services needed to make appropriate health decisions."[19] If patients are engaged on the basis of preexisting connections with investigators and health care institutions—institutions of the kind that are competitive for PCORI awards—they are probably able to navigate the health care system with comparative sophistication and ease. Findings regarding the number of college and graduate degrees held by engaged patients, detailed above, support this hypothesis; educational attainment is often used to estimate health literacy.[20]

Low health literacy is associated with differential use of health care services and health-related outcomes.[21] Hard-to-reach patients will have unique experiences, perspectives, and understandings of the barriers they face, which seem to be particularly relevant to PCORI's goal of ensuring that research will meaningfully contribute to real-world improvements in patient care. What those improvements will need to look like for savvy consumers of health care as compared to those who are more marginalized is probably substantially different. Achieving representativeness among engaged patients is, therefore, a means of combating disparities and promoting health equity.[22]

Beyond health literacy, there are a number of relevant traits that could be expected to influence the sort of input engaged patients might provide. For example, for reasons similar to those driving health literacy, patients engaged via convenience sampling are more likely to have trust in the health care system, in their clinicians, and in researchers. The majority of engaged patients interviewed by Ellis and Kass were white, and race is known to correlate with trust both in the health care system and in research.[23] Relative levels of trust will be particularly important if engaged patients are being used to gauge, for instance, how comfortable prospective study subjects will be with certain research-related risks, the acceptability of elements of study design, or what recruiting methods would be most effective. It may also skew the perceived acceptability or responsiveness of the intervention being tested. Furthermore, the types of patients that have the time and money to engage with researchers—in addition to working, caregiving, or fulfilling other obligations—are likely to be relatively better off. Preferentially including well-connected, well-informed, and well-off patients in research may perpetuate disadvantages experienced by marginalized groups.

As long as ties between patient advocacy organizations and industry remain prevalent, researchers who engage patients affiliated with PAOs may hear patient voices filtered through an industry lens.

Relying on patients who are not well trained or well-informed. PCORI-funded investigators are steeped in the Western-medicine and research paradigms. Patients may have different frameworks for thinking about health and wellness and are likely to be much less familiar (if they are familiar at all) with research methods.[24] Patient engagement will be reduced to mere tokenism if patients are not capable of meaningfully contributing to the research enterprise due to a lack of preparation and training.

In a sense, bringing individuals who cannot be effective collaborators into patient-engaged research creates the inverse of the problem described in the previous section. Choosing the "model" patient may lead to confirmation bias because investigators are engaging with patients so enmeshed in the health care system that they do not offer sufficiently distinct perspectives to be valuable. By contrast, relying on patients who may have unique, relevant perspectives to share but who lack the understanding and confidence to express those perspectives in a clear and constructive manner may result in PIs' hearing nothing at all or receiving input that is impractical or out of place. For example, patients may be hesitant to contribute or may not understand how best to contribute if the study team speaks to them in a rush of acronyms—"INDs," "IRBs," "RCTs," "DSMBs"—that are familiar to researchers but for which most patients (reasonably and predictably) have no context. In these scenarios, patient involvement becomes an exercise in "checking the box" simply to satisfy PCORI's technical requirements.[25] Without training, notes Rebecca Dresser, engaged patients "are likely to become frustrated and cynical about researchers' motives for including them,"[26] which can damage trust or have other negative effects on research, potentially causing more damage than if patients had not been engaged at all. Outsider perspectives are essential, but the outsiders must be adequately supported.

Overreliance on patient advocacy organizations. A third consideration is that patients selected for engagement at present often have close ties to patient advocacy organizations. PAOs are formally organized nonprofit groups that, as one of us (Matthew McCoy) and colleagues describe them, seek "to combat a particular disease or disability or to work toward improving the health and well-being of a particular patient

population."[27] The organizations play an influential role in shaping health policy and are obvious allies for investigators doing patient-centered outcomes research. However, PAOs often differ in systematic ways from the broader patient population. To the extent that patients affiliated with PAOs share the values and interests of their organizations, their perspectives on research are likely to reflect these differences. There are at least three ways in which the views of PAO-affiliated patients can diverge from those of other patients.

First, the broader population of persons affected by a particular disease or disability may not speak with one voice, and PAOs may champion research priorities not shared by all patients. For example, the autism community faces schisms over how to direct funding for research, and Autism Speaks, a leading autism advocacy organization, has repeatedly been criticized by outsiders and by people who have left the organization for its research priorities. In the past, the organization was criticized for continuing to support vaccine research despite mounting evidence that vaccines were not a cause of autism (although the organization is now clear that "[v]accines do not cause autism"[28]), and others have criticized its emphasis on curing autism.[29] Similar observations can be and have been made about advocacy movements around breast cancer,[30] mental health,[31] and other health issues. Ethical problems arise when groups with the best access to researchers gain a disproportionate advantage, as the imbalance promotes unfair and inequitable allocations of research funding and prioritization of some research goals at the expense of other equally valid (indeed, perhaps more valid) goals.

Second, many PAOs have well-documented financial and governance relationships with drug, device, and biotech companies. As a recent study showed, "among 104 of the largest U.S.-based [PAOs], at least 83% received financial support from drug, device, and biotechnology companies, and at least 39% [had] a current or former industry executive on the governing board."[32] These relationships give rise to institutional conflicts of interest that can influence PAOs to act in ways that benefit their industry backers at the expense of the patients they purport to serve. While there have been reports of overt pressure on industry-funded PAOs to adjust their priorities to suit donor interests, COIs need not result in this type of demonstrable pressure to raise ethical concerns.[33] Institutional COIs are a result of circumstances in which an organization's financial interests pose a risk of unduly influencing the organization's pursuit of its primary mission. Thus, a COI can exist even without the actual occurrence of undue influence.[34] Many PAOs are mindful of these conflicts and recognize that more substantial efforts to manage them are needed. For example, a 2017 survey of PAO leaders found that "most . . . believe that industry [COIs] are relevant to PAOs [and] acknowledge that their policies need to be strengthened."[35] However, as long as ties between PAOs and industry remain prevalent, researchers who engage patients affiliated with PAOs may hear patient voices filtered through that lens.

Third, while some PAOs may be biased by industry ties but still function more or less independently, some groups presenting themselves as PAOs are essentially industry fronts. "Astroturfing" is the creation of a fake grassroots organization to create the perception that there is public support for an industry agenda.[36] For example, Sprout Pharmaceuticals created what was widely regarded as a sham organization called "Even the Score" to shill for Addyi, its antidepressant-turned-female-aphrodisiac.[37] Addyi, which twice failed to get U.S. Food and Drug Administration approval, received approval (albeit with a black-box warning) after Even the Score mounted an ostensibly grassroots campaign linking Addyi's approval to women's sexual health equity. Consumer groups paid by Even the Score advocated for FDA approval, but consumer groups that were not paid by Even the Score opposed both Addyi's approval and use. Although "Female Viagra," as Addyi is sometimes called, has not been successful in the marketplace, commentators have expressed concern that the Even the Score campaign taught companies to manipulate the FDA through patient advocacy.[38] The development of Addyi is not an example of patient-engaged research, but it vividly illustrates how patient engagement can be hijacked by industry for its own ends. One could easily imagine a similar example in which companies—motivated by the obvious and growing political influence of patients—hire patients to advance

RECOMMENDATIONS

- Engage more patients—Involve larger numbers of patients to capture a wider variety of perspectives and experiences.
- Purposively engage patients—Use purposive sampling to ensure representativeness along relevant dimensions.
- Reduce barriers to engagement—Identify and address obstacles to engagement of a representative range of patients.
- Offer training to facilitate engagement—Make training on research and research methods available to any patient interested in engaging.
- Manage engaged patients' conflicts of interest—Require disclosure of COIs and consider further steps to mitigate them.
- Conduct further research on who is engaged—Determine which patients are presently engaged in research and assess their representativeness.

their agendas through collaboration with PCORI-funded investigators.

The sympathetic nature of patients imparts credibility to their recommendations about research. But the prospect of PAOs with research agendas that are not broadly shared by the patient community, of conflicted PAOs, and of sham patient organizations suggests the danger of assuming that every organization that purports to represent patient interests truly does. Instead of ensuring patients an independent voice in the development of research priorities and projects, the involvement of PAOs could be used to give industry additional avenues for promoting its agenda. Some investigators may be able to discern these issues when they arise, but it would be naïve to assume that they will be able to recognize and avoid such outside influence reliably.[39]

Recommendations

In the previous section, we identified ways in which representativeness may be compromised by current patient-engagement strategies. These shortcomings are more concerning in instances where engaged patients are granted a substantial role in shaping research. Nevertheless, we should promote diversity in patient engagement. In what follows, we offer several suggestions for more conscientious selection of patients for engagement (see the table). Some of these strategies may have cross-cutting effects and increase representativeness broadly, while others, though important, are likely to have a more limited effect.

Engage more patients. The number of patients engaged may help address the problems of nonrepresentativeness. One study found that although 15 percent of projects reported engaging only one patient, more than half of projects reported engaging six or more patients.[40] These numbers are encouraging. While every patient's experience is important, experiential knowledge is necessarily both limited and idiosyncratic;[41] as Dresser notes, "Personal experience cannot in itself confer knowledge of 'what it is like' for others in similar situations."[42] Therefore, including more patients is a first step toward collecting the desired variety of perspectives.

Purposively engage patients. Numbers alone cannot ensure that engaged patients are representative of the target patient population along all relevant dimensions. It is also necessary to use purposive—rather than convenience—sampling. The limited available evidence suggests this is not presently happening in patient-centered outcomes research. As noted above, it appears to be quite rare for research teams to recruit patients for engagement with the aim of balancing competing viewpoints or with the goal of achieving socioeconomic or other types of experiential diversity.

When patients are engaged in research, representativeness needs to be considered along a number of axes. In most cases, it is necessary to consider the typical indicators of diversity, such as gender, race, educational attainment, and socioeconomic status. Ultimately, however, representativeness must be understood contextually, in relation to the population of interest for the research. For example, breast cancer disproportionately affects women, and the majority of sickle cell patients are black. Achieving a 50-50 mix between men and women engaging with researchers about a breast cancer study is probably not desirable, just as proportioning racial diversity in accordance with U.S. census numbers is probably inappropriate in a sickle cell study. Representativeness should also account for heterogeneity in disease progression or along the spectrum of illness—such as the autism spectrum, which captures a wide range of conditions—because individuals may reasonably be expected to have different interests depending on where they fall on the continuum. It is also important to recruit from subpopulations that are often underrepresented in research, such as people sixty-five and older.[43]

Further, investigators have an obligation to understand reasonable debates and disagreements within the relevant patient community and to engage with patients embodying these diverse views. For example, members of the Alzheimer's community have different views on the relative importance of cure versus caregiver support.[44] But it is not necessary to give credence to unmeritorious viewpoints in an attempt to achieve a veneer of balance. It would be inappropriate, for instance, to engage antivaxxers in research on the causes of autism, given the current state of the science.

Investigators have an obligation to understand reasonable debates and disagreements within the relevant patient community and to engage with patients embodying these diverse views.

PCORI has sought to overcome barriers to patient engagement in research. For example, PCORI's Pipeline to Proposal awards program focuses on building the community of patients and other stakeholders who can participate in patient-centered outcomes research by providing funding to build capacity and engage people around health care research.[45] It would be useful if these funds were awarded with a preference for inclusion of diverse patient perspectives, as with university research grant programs that seek to promote interdisciplinary work by conditioning funding on the inclusion of faculty

members from different university departments and schools.

As a further step, PCORI should ask all funded investigators to report how patients were chosen for engagement and why. PCORI currently requests that funding proposals include "a plan to ensure representativeness of [research] participants."[46] PCORI should request a similar plan for patient engagement.

Reduce barriers to engagement. Serving as an engaged patient will impose opportunity costs for everyone. Barriers to participation are, however, likely to be higher for some patients than for others. Because these barriers are not randomly distributed, they can be expected to affect representativeness.

Offering payment to engaged patients could address a financial barrier to engagement for some individuals. PCORI offers a compensation framework that emphasizes the importance of fair financial compensation as a means of recognizing the contributions of all members of the research team, including patients.[47] Such acknowledgment is an important function of payment.[48] Yet payment may have an additional benefit here if it expands the pool of patient partners who are willing and able to engage. It may also be necessary to hold meetings during evenings and weekends, or whenever more patients are able to come, and to find other ways to be flexible, such as using conference calls for patients who may be too ill to reliably attend in-person meetings or ordering rides for participants via ride-sharing apps.

Offer training to facilitate engagement. To be effective research partners, engaged patients will need some understanding of the science and the research process. Training may, therefore, appear to be part of the "how" of patient engagement, a question we have set to the side in this article. Yet we would argue that, unless training is made readily available for all engaged patients, researchers will continue to rely disproportionately on the most well-educated and well-informed patients, creating the risks of confirmation bias discussed above.

Theoretically, a number of stakeholders in the research could offer such training. There are barriers to having PCORI-funded investigators do it, however, such as lack of time and resources, the inefficiency in having every PI reinvent the wheel to develop and offer training, and the possibility that the investigators might, even if unintentionally, impart knowledge that conforms with their views, diluting the value of patient engagement. At present, some patient groups are able to provide their members with the necessary education. For instance, the Parkinson's Disease Foundation has a Parkinson's Advocates in Research program that has trained more than 280 volunteer patient advocates "to work on the frontlines with the professionals seeking better . . . treatments" for Parkinson's disease since 2008.[49] The foundation explains that, when advocates from disease communities "are primary partners in research alongside scientists, industry, and government," research can be made more efficient and effective. Some patients cannot, however, access training, and some organizations cannot offer it. For these individuals and groups, reliance on an ad hoc system of training may limit meaningful participation—or perhaps preclude any participation—resulting in marginalization. The gold standard should therefore be training that is available to any patient and delivered by an institutional stakeholder.

PCORI is an obvious candidate for developing and delivering this information, even recognizing that PCORI-funded projects are heterogeneous and that a PCORI-developed program may not be perfectly tailored to all projects' needs. PCORI would benefit from economies of scale and from its own considerable institutional knowledge. Further, PCORI is already taking steps in this direction.[50] For example, the PCORI Ambassador program "equips, trains, connects, and mobilizes patients, organizations, and other stakeholders to share PCORI's vision and mission, . . . participate as full partners in research, and help ensure the sharing and use of information generated from PCORI-funded projects."[51] All ambassadors, who are volunteers, must complete PCOR 101 and PCOR Science Training and fill out a COI disclosure form, although mechanisms for subsequent conflict management are unclear. At the end of 2014, PCORI had trained eighty-two ambassadors from stakeholder communities—not just patients but also caregivers, researchers, clinicians, purchasers, payers, industry representatives, and others.[52] The long-term goal is to have ambassadors in every state. This is an important effort but of limited effectiveness given the small reach of the program at present.

A longer-term but nonetheless salient concern associated with training is that it may turn engaged patients into "insiders," thereby depriving them of the "outsider" perspective that is implicitly their contribution to patient-centered outcomes research. If one's expertise depends on a relative lack of knowledge about the nature and details of research, does acting as an engaged patient undermine the patient's "outsider" expertise over time? Comparisons can be drawn to the community member on an institutional review board—a lay person who often fulfills the regulatory roles of a nonscientific member and unaffiliated member. The longer these individuals are exposed to IRB deliberations, the less likely they are to act as outsiders.[53] U.S. regulations do not require the IRB's community member to be representative of the community, however; the concept of patient-engaged research does demand representativeness and suggests that participation as an engaged patient may need to be time limited. Rather than engaging the same patients on multiple studies and over time, researchers, with the assistance of PCORI, should strive for reasonable turnover in the population of engaged patients.

Manage engaged patients' conflicts of interest. Many see the involvement of PAOs in PCORI-funded research as making a valuable contribution to research rather than as compromising it. And given that PAOs often unite many patients under one umbrella, these organizations may indeed offer important perspectives. To maximize these benefits, however, it is important to actively manage the COIs previously described.

Disclosure of COIs has become a widely accepted norm in science and medicine. Engaged patients, who are being held up as important contributors to research, should be held to the same standards as researchers in this regard. Thus, it is important that patients disclose both individual COIs and the COIs of organizations with which they are affiliated. This two-step disclosure is important so that it is possible to identify when patients are affiliated with an industry-funded advocacy organization. Requiring disclosure is not the same as assuming that PAOs would knowingly let their financial ties bias their judgment. Rather, requiring disclosure is an effort to achieve transparency and promote trustworthiness.[54]

Disclosure is not a complete solution to COIs. Compelling engaged patients to provide full information about their COIs is, however, a first step to enabling additional solutions to ensure that industry interests do not unduly influence patient engagement. A further step would be to limit the engagement of PAO-affiliated patients to those from PAOs that receive less than a certain percentage, perhaps 10 percent, of their total funding from industry. Limiting industry funding in this way is a means of splitting the difference between prohibiting PAO involvement (which is likely a nonstarter and would sacrifice some benefits) and allowing egregious cases in which PAOs become de facto advocates for industry. A further step would be to cap the number of engaged patients who are involved in industry-funded PAOs in a given study.

Conduct further research on who is engaged. Finally, more research is needed to determine which patients are being engaged. Ideally, to assess the full extent to which the potential pitfalls discussed here are encountered in practice, it would be helpful to gather more individual-level information about engaged patients and also more information about how researchers identify these patients for engagement and why they select some patients over others. In addition, PCORI should continue funding research on patient engagement, with a focus on how to effectively recruit patients from hard-to-reach populations.

Taking Patient Engagement Seriously

PCORI aims to increase the quality of information available to patients in their health-related decision-making, both through its own funding and by influencing health care research funded by others to be more patient-centered. Moreover, there is a general trend toward patient engagement in health-related decision-making.[55] Patient-centered outcomes research is therefore a useful case study for drawing lessons about high-quality patient engagement in research that can apply more broadly. Even if the problems we have identified are infrequent, the obvious potential for bias, injustice, and the erosion of public confidence in research makes it important that PCORI and other bodies supporting patient engagement create robust policies to address them. If we are going to take patient engagement seriously as a goal, we need to take seriously the question of who the patients are and ought to be.

Notes

1. Patient Protection and Affordable Care Act, 2010.
2. "About Us," Patient-Centered Outcomes Research Institute, March 21, 2017, https://www.pcori.org/about-us.
3. "Our Story," Patient-Centered Outcomes Research Institute, August 5, 2014, https://www.pcori.org/about-us/our-story.
4. "How We Select Research Topics," Patient-Centered Outcomes Research Institute, May 1, 2014, https://www.pcori.org/research-results/how-we-select-research-topics.
5. E. A. Largent et al., "Patient-Centered Outcomes Research: Stakeholder Perspectives and Ethical and Regulatory Oversight Issues," *IRB: Ethics & Human Research* 40, no. 1 (2018): 7-17.
6. L. Esmail, E. Moore, and A. Rein, "Evaluating Patient and Stakeholder Engagement in Research: Moving from Theory to Practice," *Journal of Comparative Effectiveness Research* 4, no. 2 (2015): 133-45.
7. L. Frank et al., "Conceptual and Practical Foundations of Patient Engagement in Research at the Patient-Centered Outcomes Research Institute," *Quality of Life Research* 24, no. 5 (2015): 1033-41.
8. S. R. Morain et al., "Deliberative Engagement Methods for Patient-Centered Outcomes Research," *The Patient—Patient-Centered Outcomes Research* 10, no. 5 (2017): 545-52.
9. J. V. Selby, A. C. Beal, and L. Frank, "The Patient-Centered Outcomes Research Institute (PCORI) National Priorities for Research and Initial Research Agenda," *Journal of the American Medical Association* 307, no. 15 (2012): 1583-84.
10. "What We Mean by Engagement," Patient-Centered Outcomes Research Institute, August 12, 2014, https://www.pcori.org/engagement/what-we-mean-engagement.
11. L. E. Ellis and N. E. Kass, "How Are PCORI-Funded Researchers Engaging Patients in Research and What Are the Ethical Implications?," *AJOB Empirical Bioethics* 8, no. 1 (2017): 1-10.
12. Ibid., 3.
13. Ibid., 5.
14. L. P. Forsythe et al., "Patient and Stakeholder Engagement in the PCORI Pilot Projects: Description and Lessons Learned," *Journal of General Internal Medicine* 31, no. 1 (2016): 13-21.
15. Ellis and Kass, "How Are PCORI-Funded Researchers Engaging Patients in Research and What Are the Ethical Implications?," 8.
16. "National Priorities and Research Agenda," Patient-Centered Outcomes Research Institute, May 1, 2014, https://www.pcori.org/research-results/research-we-support/national-priorities-and-research-agenda.
17. D. Cukor et al., "Patient and Other Stakeholder Engagement in Patient-Centered Outcomes Research Institute Funded Studies of Patients with Kidney Diseases,"

Clinical Journal of the American Society of Nephrology 11, no. 9 (2016): 1703-12.

18. J. V. Selby, L. Forsythe, and H. C. Sox, "Stakeholder-Driven Comparative Effectiveness Research: An Update From PCORI," *Journal of the American Medical Association* 314, no. 21 (2015): 2235-36.

19. Institute of Medicine, *Health Literacy: A Prescription to End Confusion* (Washington, D.C.: National Academies Press, 2004).

20. L. D. Chew, K. A. Bradley, and E. J. Boyko, "Brief Questions to Identify Patients with Inadequate Health Literacy," *Family Medicine* 36, no. 8 (2004): 588-94.

21. N. D. Berkman et al., "Low Health Literacy and Health Outcomes: An Updated Systematic Review," *Annals of Internal Medicine* 155, no. 2 (2011): 97.

22. R. Hasnain-Wynia and A. C. Beal, "Role of the Patient-Centered Outcomes Research Institute in Addressing Disparities and Engaging Patients in Clinical Research," *Clinical Therapeutics* 36, no. 5 (2014): 619-23.

23. L. E. Boulware et al., "Race and Trust in the Health Care System," *Public Health Reports* 118, no. 4 (2003): 358-65.

24. M. Tai-Seale et al., "The Language of Engagement: 'Aha!' Moments from Engaging Patients and Community Partners in Two Pilot Projects of the Patient-Centered Outcomes Research Institute," *Permanente Journal* 20, no. 2 (2016): 89-92.

25. Largent et al., "Patient-Centered Outcomes Research."

26. Rebecca Dresser, *When Science Offers Salvation: Patient Advocacy and Research Ethics* (Oxford University Press, 2001), 41.

27. M. S. McCoy et al., "Conflicts of Interest for Patient-Advocacy Organizations," *New England Journal of Medicine* 376 (2017): 880-85, at 880.

28. "What Causes Autism?," Autism Speaks, July 25, 2012, https://www.autismspeaks.org/what-autism/learn-more-autism/what-causes-autism.

29. E. Shire, "'Autism Speaks'—but Should Everyone Listen?," *Daily Beast*, June 13, 2014.

30. P. Orenstein, "Our Feel-Good War on Breast Cancer," *New York Times*, April 25, 2013.

31. N. Tomes, "The Patient as a Policy Factor: A Historical Case Study of the Consumer/Survivor Movement in Mental Health," *Health Affairs* 25, no. 3 (2006): 720-29.

32. McCoy et al., "Conflicts of Interest for Patient-Advocacy Organizations," 884.

33. K. Thomas, "Furor over Drug Prices Puts Patient Advocacy Groups in Bind," *New York Times*, September 27, 2016.

34. M. S. McCoy and E. J. Emanuel, "Why There Are No 'Potential' Conflicts of Interest," *Journal of the American Medical Association* 317, no. 17 (2017): 1721-22.

35. S. L. Rose et al., "Patient Advocacy Organizations, Industry Funding, and Conflicts of Interest," *JAMA Internal Medicine* 177, no. 3 (2017): 344-50, at 348.

36. S. Perry, "New Patient-Advocacy Group 'Outed' by Minnesota-Based Website as 'Astroturf' Campaign," *MinnPost*, February 16, 2016, https://www.minnpost.com/second-opinion/2016/02/new-patient-advocacy-group-outed-minnesota-based-website-astroturf-campaign.

37. A. Hogenmiller, A. Hirsch, and A. Fugh-Berman, "The Score Is Even," *Bioethics Forum* (blog), June 14, 2017, https://www.thehastingscenter.org/the-score-is-even/.

38. J. Block and L. Canner, "The Grassroots Campaign for 'Female Viagra' Was Funded by Its Manufacturer," *The Cut*, accessed June 14, 2018, https://www.thecut.com/2016/09/how-addyi-the-female-viagra-won-fda-approval.html.

39. E. Silverman, "McCaskill Bill Would Make Pharma Report Payments to Patient Advocacy Groups," *STAT*, June 6, 2018, https://www.statnews.com/pharmalot/2018/06/06/mccaskill-payments-advocacy-groups/.

40. Forsythe et al., "Patient and Stakeholder Engagement in the PCORI Pilot Projects."

41. G. P. Martin, "'Ordinary People Only': Knowledge, Representativeness, and the Publics of Public Participation in Healthcare," *Sociology of Health & Illness* 30, no. 1 (2008): 35-54.

42. R. Dresser, "The Role of Patient Advocates and Public Representatives in Research" in *The Oxford Textbook of Clinical Research Ethics*, ed. E. J. Emanuel et al. (New York: Oxford University Press, 2011), 234.

43. K. S. Kauffman et al., "Engaging Hard-to-Reach Patients in Patient-Centered Outcomes Research," *Journal of Comparative Effectiveness Research* 2, no. 3 (2013): 313-24.

44. J. Graham, "Alzheimer's Activists Splinter in Angry Fight over Priorities," *STAT*, March 2, 2016.

45. "Pipeline to Proposal Awards," Patient-Centered Outcomes Research Institute, August 5, 2014, https://www.pcori.org/funding-opportunities/research-support-funding-opportunities/pipeline-proposal-awards.

46. "PCORI Methodology Standards," Patient-Centered Outcomes Research Institute, November 12, 2015, at https://www.pcori.org/research-results/about-our-research/research-methodology/pcori-methodology-standards.

47. "Financial Compensation of Patients, Caregivers, and Patient/Caregiver Organizations Engaged in PCORI-Funded Research as Engaged Research Partners," Patient-Centered Outcomes Research Institute, June 10, 2015, www.pcori.org/sites/default/files/PCORI-Compensation-Framework-for-Engaged-Research-Partners.pdf.

48. L. Gelinas et al., "A Framework for Ethical Payment to Research Participants," *New England Journal of Medicine* 378 (2018): 766-71.

49. "Become a Research Advocate," Parkinson's Foundation, October 12, 2017, http://parkinson.org/research/Patient-Engagement/Advocate-for-Research.

50. L. Gelinas et al., "Oversight of Patient Centered Outcomes Research: Recommendations from a Delphi Panel," *Annals of Internal Medicine* (forthcoming).

51. "Pipeline to Proposal Awards," Patient-Centered Outcomes Research Institute.

52. L. Frank et al., "Conceptual and Practical Foundations of Patient Engagement in Research at the Patient-Centered Outcomes Research Institute," *Quality of Life Research* 24, no. 5 (2015): 1033-41.

53. C. W. Lidz et al., "The Participation of Community Members on Medical Institutional Review Boards," *Journal of Empirical Research on Human Research Ethics* 7, no. 1 (2012): 1-8.

54. S. L. Rose, "Patient Advocacy Organizations: Institutional Conflicts of Interest, Trust, and Trustworthiness," *Journal of Law, Medicine & Ethics* 41, no. 3 (2013): 680-87.

55. "Learn about FDA Patient Engagement," U.S. Food and Drug Administration, accessed August 9, 2018, https://www.fda.gov/ForPatients/PatientEngagement/default.htm.

NOTES FROM SESSION 4