



Engaging People With ME¹ As Partners in the Collaborative Research Centers

For the first time in more than ten years, the National Institutes of Health has issued Requests for Applications (RFA) for research into ME.² The RFA for the Collaborative Research Centers requires that each Center have “a plan for outreach and partnering with other ME/CFS stakeholders.” The RFA also states, “Applicants are strongly encouraged to establish partnerships with patients groups and solicit their input on recruitment and the clinical meaningfulness and design of the question under study.”

In this document, we share suggested best practices for engaging people with ME as true partners in the work of the Collaborative Research Centers. These recommendations are based on selected literature on patient engagement, particularly from the Patient-Centered Outcomes Research Institute (PCORI). The co-authors of this document have been research subjects, health care practitioners, and/or patient representatives in a variety of contexts. While this document focuses on engaging people with ME, caregivers and family members, and advocacy organizations, there are other stakeholders that should also be taken into account. We hope this document will assist applicants in designing community engagement plans tailored to their Centers.

I. Why Engagement is Important

We, the community of people with ME, honor and respect the researchers who choose to study this complex disease. You are developing the fundamental building blocks on our path to treatments and ultimate recovery. You are our lifeline.

This debilitating disease destroys lives in many ways. Preliminary data suggest that ME may increase the risk of heart disease and some cancers, as well as death from suicide. But ME also kills our ability to pursue our passions, to interact with our families, to be productive members of society, and to fully live our lives. We deserve better.

Your research will help determine if we are able to return to our friends in school and live a normal teen life, return to college or work, raise our children as full participants in their lives, and play with and help care for our grandchildren. We need your help to once again be productive members of society and to create lives that match our dreams.

¹ We are using ME in this document due to the preference of most in the community although the Requests for Applications referenced in this document refer to our disease as “ME/CFS”

² <https://grants.nih.gov/grants/guide/rfa-files/RFA-NS-17-021.html>
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People with ME need you. However, you need us as well. As Patient Partners, we respect the answer of “I don’t know...yet”. We don’t have all the answers, but we do know our bodies and our lives. With a seat at the table, we can provide you with insights and perspectives you may not have previously considered. We can help you understand the diverse perspectives in our community, and we can leverage our expertise to improve your research projects.

We are excited to share some Patient Partner best practices to facilitate a deeper relationship, which will only work to enhance the research. It is important to note that these best practices do not represent a one size fits all approach. Every research project is different, and therefore the best ways to engage Patient Partners will vary. Rather, these best practices are better viewed as tools in a toolbox. You and your Patient Partners will be best suited to select the tools that fit your research project.

People with ME are so passionate about contributing to the research that many are willing to make deep personal and physical sacrifices. As Patient Partners attend your meetings and provide insights, you will get to know us outside the formal office visit or lab setting. You may see us struggle through brain fog or become fatigued when engaged in conversation too long. You will see a glimpse of what our inner circle witnesses on a daily basis. However, this must not deter our joint efforts to enhance the research by having all perspectives at the table. We know the risks and potential toll it takes to be active Patient Partners, and we are willing to make that sacrifice. Although many of us will require special accommodations to participate as Partners, recognition that we have something to offer all phases of ME research will only help advance this cause.

As people living with ME, we are excited to partner with you as equals through the entirety of your research lifecycle. There should be “Nothing about me, without me.”

II. **General Principles of Patient Partner Engagement**

Definition of Terms

In the most general terms, a stakeholder is a person with an interest or concern in something. Indeed, in reviewing the literature on engaging patients in medical research, we found that the term “stakeholder” is used quite often. However, there is no standardized usage of terms like stakeholders, patients, subjects, community, and partners, so we recommend the following terms for the sake of clarity.

According to PCORI Engagement Rubric for Applicants, from the Patient-Centered Outcomes Research Institute:

- “Patient partners’ is intended to include patients (those with lived experience), family members, caregivers, and the organizations that are representative of the population of interest in the particular study.

- It is important that patient partners are not confused with patient subjects; patient partners are members of the research team and involved in the planning, conduct, and dissemination of the research, whereas patient subjects are those individuals actually enrolled into the study as participants.
- ‘Stakeholder partners’ may include members of constituencies based on professional, rather than personal, experience. For example, these constituencies can include: clinicians, purchasers, payers, industry, hospitals and health systems, policy makers, and training institutions. Some individuals may fit into several categories.”³

As the people who suffer the range of debilitating symptoms of ME every day, we have the greatest interest and investment in the outcomes of ME research. The new Collaborative Research Centers will need to consider the full spectrum of stakeholder partners, but people with ME should be at the center as both subjects and Patient Partners.

Just as there are diverse groups of stakeholders, there is great diversity among people with ME. Meaningful, substantive engagement should capture that diversity as much as possible. To be robust, engagement with Patient Partners is not a one time thing. The RFA strongly recommends our input be sought “on recruitment and the clinical meaningfulness and design of the question under study.” Systematically incorporating our input on these fundamental questions will require sustained engagement over time, supported by strong internal processes and allocation of resources.

Values

We have found the conceptual model underlying patient centered outcomes research to be helpful when thinking about how to ensure people with ME are at the center of all research. The principles of patient-centered research include trust, honesty, co-learning, transparency, reciprocal relationships, and partnership. Awardees of PCORI research grants have reported that when these principles are used, meaningful Patient Partner participation can be enhanced. More specific contributions made by engaged Partners have included “changes to project methods, outcomes or goals, modifications to interventions, improvement of measurement tools and data collection methods, and interpretation of qualitative data.”⁴

In order to ensure that Patient Partners represent a wide range of the community, it is important that recruitment announcements for Patient Partners be made in a diverse set of publications and patient-focused websites and that the recruitment process be as transparent and unbiased as possible. The goal should be to recruit Patient Partners (both people with ME and caregivers) of varying ages, disease severity, races, sexual orientations, ethnicities, and locations. Due to the nature of ME you will need to plan how to offer accommodations for the fluctuating symptoms encountered by people with ME; details on what to keep in mind are discussed later in this document.

³ PCORI Engagement Rubric 2015.

⁴ Frank et al. 2015.

As discussed in a PCORI study on engaging hard-to-reach patients:⁵ “in order to make patient-centered outcomes research more meaningful to patients and their caregivers, patient-centered outcomes research should be conducted with a focus on building and maintaining trust, which is achieved via pre-engagement with communities and continuous engagement of study participants and their communities.”⁶ Patient Partners can offer more value in a research project when their role is perceived to be genuine and not simply symbolic.

A number of case studies in the literature emphasize the iterative nature of Patient Partner engagement (See Section IV, Additional Resources). Co-learning and reciprocal relationships can help avoid or solve miscommunications. Neither researchers nor Patient Partners are experts in full engagement, and missteps will happen. No one will get this right the first time, but we can make progress together if we cultivate an attitude of mutual learning. This responsibility is shared by researchers and Partners alike.

There is a lot of understandable distrust in the ME community due to decades of disrespect and mistreatment from some researchers, medical professionals, and funding agencies. Some people with ME are unwilling to publicly disclose their diagnoses, and others are still navigating the path to diagnosis. There are also strong differences of opinion among people with ME, researchers, and funders over a variety of scientific issues and priorities. These tensions make it all the more vital that these principles be kept in mind at all times in order for trust to be earned and maintained among everyone involved.

Compensation

Compensation for work done, with the exception of the occasional volunteer and intern, is generally expected for all types of labor. The business of research is no different. Patient subjects in research trials are often compensated monetarily or by receiving free related care. The same should be true for Patient Partners.

PCORI notes that: “Fair financial compensation demonstrates that patients, caregivers, and patient/caregiver organizations’ contributions to the research, including related commitments of time and effort, are valuable and valued. Compensation demonstrates recognition of the value, worth, fairness of treatment with others involved in the research project, and contributes to all members of the research team being valued as contributors to the research project.”⁷ There is an informative report⁸ on a PCORI research study that goes into detail on how two patients were incorporated into the project as full partners and compensated accordingly.

⁵ Note that many people with ME are “hard to reach” because they frequently are housebound and/or not able to see the few experts who diagnose and treat the disease.

⁶ Kauffman et al. 2013.

⁷ PCORI Compensation Framework 2015.

⁸ Robbins et al 2016.

Just as researchers and their team of medical professionals, administrators and statisticians receive payment for their efforts, Patient Partners who aid in the advancement of medical research should also receive fair and equitable compensation based on their time, expertise, responsibility, type of participation, and overall contributions to the advancement of the specific study. Patient Partners who share in decision-making may be compensated on par with other research team members, while those who simply share information might be offered in-kind vouchers as payment. In all cases, however, compensation extends beyond out-of-pocket expenses and fairly reflects Patient Partners' skills and contributions.

How to compensate Patient Partners is a more complex question than one may realize. Obviously, compensation should comply with local, state and federal tax laws and wage requirements. In addition, many people with ME are receiving disability benefits, and there are limits on the amount of income a person can earn without jeopardizing those benefits. Disability insurance companies routinely examine the online and work-related activities of beneficiaries, and this can easily escalate into a dispute over the significance of those activities. Individual circumstances vary widely, and these issues should be considered and discussed with your Patient Partners in advance. Alternative forms of compensation, such as gift cards, may be appropriate for some individuals.

Attention should also be paid to the costs associated with Patient Partners' involvement. Video conferencing, travel to meetings, and other required expenses should be considered in advance and included in the research budget. Your Patient Partners will incur other financial costs as well, such as public transportation to attend meetings, parking, childcare arrangements, meals, and supplies. The best practice here is to discuss these issues proactively with both your potential and engaged Patient Partners. Be flexible in defining fair compensation that meets their needs, and be sure to include this as a financial line item in your budget.

Accommodations

Patient Partner engagement in research is in its infancy, and there is a learning curve for both researchers and partners alike. People living with ME experience a wide range of severity of symptoms, and this diversity offers great potential insights to researchers. However, that range of severity means that a variety of accommodations may be necessary to facilitate the effective engagement of Patient Partners. This is no different from working with people with any type of disability.

The specifics of how to accommodate Patient Partners will vary depending on the Partners' personal circumstances, range and severity of symptoms, as well as the type, scope, duration and location of the research project itself. Examples of accommodations include, but are not limited to:

- Assistance with transportation to and from in-person meetings
- Virtual meeting attendance via video conferencing
- Accommodations for wheelchairs, rollators and other mobility aids

- Limits on meeting length and frequency; perhaps more frequent breaks
- Extra time to review materials and prepare for meetings
- Flexibility in accommodating fluctuating symptoms
- Ways to provide input and feedback electronically and/or outside normal business hours
- Fragrance free space; modifications to lights and sound
- Hydration and positional modifications

People with ME are willing to partner with researchers, despite the ramifications our bodies may endure. But accommodations will make this easier, by protecting our bodies and enabling severely affected people to more fully participate. Because of the diversity of our experiences, the best advice is to ask your Patient Partners what would make it easier for them to participate. It can be hard for people with ME to be honest about the accommodations they need, partially because so few accommodations are offered them. Accessibility does not equal ramps. Talk to your Patient Partners, and continue the conversation throughout the lifecycle of your research.

III. Engagement Throughout the Research Cycle

Planning the Study

Working with Patient Partners as early as possible is critically important to the overall success of the study. Partner involvement during the beginning phases of the research design allows for their input on key issues, such as the questions that need to be asked, the necessary outcomes of the research, and identifying the best subject pool.

We recommend that there be a clear point of contact for Patient Partners on your team who has the resources and knowledge to answer questions and provide assistance. Since people with ME can have their health go downhill unexpectedly, it is important to discuss in advance what will happen should people have to take time off or leave the project.

Researchers should communicate early on and frequently with Patient Partners, patient advocates and other stakeholder partners in order to consider and address the ideas and priorities of these diverse groups. Forms of communication could include face-to-face, phone, email, video calling, handwritten notes, etc. During this phase you should also decide about the incentives and support you plan to offer your subjects, and should budget accordingly. These incentives could come in various forms. Subjects are sacrificing a lot to be a part of your study and that should be taken into account by the research team. As discussed earlier, compensation should also be budgeted for Patient Partners based on their responsibilities, qualifications and needs.

We recognize that Patient Partner involvement as described in these best practices is a new concept for both researchers and people with ME. Researchers and Partners will both need training and practice to work together in this way. The expectations for involvement should be

discussed and clearly articulated. Many academic institutions have resources to help facilitate Partner involvement, and these should be leveraged wherever possible.

Conducting the Study

There are many ways that your Patient Partners can continue to assist you during the conduct of your study (See Section IV, Additional Resources). Partners can help with recruitment, monitoring data collection, monitoring subject safety, devising retention strategies, and troubleshooting. New questions and hypotheses can be generated based on preliminary data. Partners can help analyze data and interpret findings, including the identification of real-world explanations or contexts for results.

Disseminating Results

The results of your research should be shared with many stakeholder groups, not just people with ME or researchers. We encourage you to consider publication in open access journals because, in the absence of access to expert care, so many people with ME must do their own literature searches. It is also critical that results be easily accessible to primary care health practitioners, since they are usually the first point of contact for people when they become sick with ME. Patient Partners can assist you in a number of ways, including drafting plain language summaries and press releases, generating media coverage, and identifying additional outlets. Depending on the study, it may be appropriate to communicate with primary care providers, news outlets, social media, alternative health care providers, schools and colleges, community groups, and ME-specific outlets such as #MEAAction, blogs and forums.

Evaluation

We strongly encourage a fluid approach to the process of advancement and learning. Lessons learned with Patient Partners should, like all research findings, be applied and incorporated into future efforts. Honest and open communication will help identify ways to expand and improve engagement with Patient Partners, as well as other stakeholders.

Engaging people with ME and other stakeholder partners will not be a short or easy task, but it is worthwhile and necessary. We need your expertise and your research more desperately than you may realize. However, our expertise can benefit your research in many ways. While the RFAs may be the current impetus for engaging with stakeholders, we hope that this will catalyze something more. People with ME can offer so much more than feedback. We can make your research more applicable, more efficient, and more successful. Let's establish partnerships, because we will make more progress together.

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IV. **Additional Resources**

1. Anderson M and McCleary K. From passengers to copilots: Patient roles expand. *Sci Transl Med.* 2015; 7:291fs25.
2. CTSA Consortium. Principles of community engagement (2nd ed.). Washington: US Government Printing Office. NIH publication #11-7782, 2011.
3. Curtis P, Slaughter-Mason S, Thielke A, Gordon C, Pettinari C, Ryan K, Church B, King V. PCORI Expert Interviews Project: Final report. Portland, OR: Center for Evidence-based Policy, Oregon Health & Science University. 2012.
<http://www.pcori.org/assets/pdfs/Expert%20Interviews%20Part%201.pdf>. Accessed March 27, 2017.
4. Darling M, Gonzalez F, Graves K, Sheppard V, Hurtado De Mendoza A, Leventhal K, Caicedo L. Practical Tips for Establishing Partnerships With Academic Researchers: A Resource Guide for Community-Based Organizations. *Prog Community Health Partnersh.* 2015 Summer;9(2):203-12.
5. Elwood, W. 2016. Community-based research: An overview [Powerpoint slides]. 12th International IACFS/ME Research and Clinical Conference: Emerging Science and Clinical Care.
6. EULAR. Patient Involvement in Research: A way to success. Zurich, European League Against Rheumatism. 2013.
http://www.eular.org/myUploadData/files/Reference_cards_explained_Booklet_pages_23-08-13_1.pdf. Accessed March 27, 2017.
7. Frank L, Forsythe L, Ellis L, Schrandt S, Sheridan S, Gerson J, Konopka K, Daugherty S. Conceptual and practical foundations of patient engagement in research at the patient-centered outcomes research institute. *Qual Life Res.* 2015;24:1033-1041.
8. Horowitz C, Robinson RMR, Seifer S. Community-Based Participatory Research from the Margin to the Mainstream: Are Researchers Prepared? *Circulation.* 2009 May 19; 119(19): 2633-2642.
9. INVOLVE (2012). Briefing notes for researchers: involving the public in NHS, public health and social care research. INVOLVE, Eastleigh. http://www.invo.org.uk/wp-content/uploads/2014/11/9938_INVOLVE_Briefing_Notes_WEB.pdf . Accessed March 27, 2017.
10. INVOLVE (2012). Developing training and support for public involvement in research. INVOLVE, Eastleigh. <http://www.invo.org.uk/wp-content/uploads/2012/11/INVOLVETrainingSupport2012.pdf>. Accessed March 30, 2017.

11. Kauffman K, dosReis S, Ross M, Barnet B, Onukwugha E, Mullins CD. Engaging hard-to-reach patients in patient-centered outcomes research. *J Comp Eff Res*. 2013 May;2(3):313-24.
12. Lavalley, DC. 2013. Enhancing Research through Patient Engagement [PowerPoint slides].
<http://www.brighamandwomens.org/Research/centers/PCERC/documents/09.23.13%20PCERC%20PE%20Workshop%20Full%20Slideshow.pdf>. Accessed March 27, 2017.
13. Mullins CD, Abdulhalim AM, Lavalley DC. 2012. Continuous Patient Engagement in Comparative Effectiveness Research. *JAMA*. 2012; 307(15):1587-588.
14. PCERC. Patient Engagement Best Practices Resource Document. Patient-centered comparative effectiveness research center, Brigham and Women's Hospital. 2013.
<http://www.brighamandwomens.org/Research/centers/PCERC/documents/PCERC%20Patient%20Engagement%20Best%20Practices%20Resource%20Document.pdf>. Accessed March 20, 2017.
15. Patient-Centered Outcomes Research Institute (Producer). 2017. Community Engagement in Research: Practical Tips for Researchers and Community Based Organizations [Video webinar]. <http://events.meetingbridge.com/join/?08120096433> Accessed March 27, 2017.
16. PCORI Compensation Framework. PCORI (Patient-Centered Outcomes Research Institute) website. <http://www.pcori.org/sites/default/files/PCORI-Compensation-Framework-for-Engaged-Research-Partners.pdf>. Published June 10, 2015. Accessed March 26, 2017.
17. PCORI Engagement Rubric. PCORI (Patient-Centered Outcomes Research Institute) website. <http://www.pcori.org/sites/default/files/Engagement-Rubric.pdf>. Published 4 Feb. 2014. Updated October 12, 2015. Accessed 27 Mar. 2017.
18. Robbins M, Tuft J, Hsu C. Learning to "Swim" with the Experts: Experiences of Two Patient Co-Investigators for a Project Funded by the Patient-Centered Outcomes Research Institute. *Perm J*. 2016 Spring; 20(2):85-88.
19. UNAIDS. Good participatory practice guidelines for biomedical HIV prevention trials. Geneva, Joint United Nations Programme on HIV/AIDS, 2011.
<http://www.avac.org/resource/good-participatory-practice-guidelines-biomedical-hiv-prevention-trials-second-edition>. Accessed March 29, 2017.
20. Woolf S, Zimmerman E, Haley A, Krist A. Authentic Engagement of Patients and Communities Can Transform Research, Practice, And Policy. *Health Affairs*. 2015;35(4):590-594.