



Involving Patients in Health Economics Research: “The PACTS Principles”

Annie Hawton^{1,2} · Kate Boddy² · Rebecca Kandiyali^{3,4} · Lynn Tatnell⁵ · Andy Gibson⁶ · Elizabeth Goodwin¹

Accepted: 19 September 2020 / Published online: 12 October 2020
© Springer Nature Switzerland AG 2020

Abstract

Discussion of public and patient involvement (PPI) in health economics (HE) research is growing. There is much literature on PPI principles and standards, but little specifically regarding involving patients in HE research. Here, we outline “PACTS”, a set of principles, developed with a PPI group, for considering patient involvement in HE research. *Planning*: Involvement is best built in to research plans from the outset. This includes setting specific goals for involvement activities, and clearly communicating the background and purpose of involvement. *Approach selection*: We describe two main approaches to involvement—discussion-based and task-based. Discussion-based approaches are useful for generating broad insights and revealing “unknown unknowns”. Task-based approaches offer a more focused means of shedding light on “known unknowns”. *Continuous involvement*: Involving patients throughout the research process and across a range of projects helps build expertise for patients and insight for HE researchers. *Team building*: Meaningful involvement creates a shared sense of ownership of the research and, over time, helps to develop a team ethos, enhancing the positive impacts of involvement. *Sensitivity*: HE research can be perceived as technical and impersonal. Addressing this requires sensitivity, clarity, and an honest and open approach. There is increased recognition that patient contributors are experts at providing a “lived experience” perspective, in the way that clinicians are experts at providing an overview of conditions and HEs are experts in the methodology of their discipline. We hope these “PACTS Principles” complement existing PPI approaches and provide a useful foundation for health economists considering patient involvement.

Electronic supplementary material The online version of this article (<https://doi.org/10.1007/s40271-020-00461-4>) contains supplementary material, which is available to authorized users.

✉ Annie Hawton
a.hawton@exeter.ac.uk

- ¹ Health Economics Group, University of Exeter Medical School, University of Exeter, Exeter, Devon, UK
- ² NIHR Applied Research Collaboration (ARC) South West Peninsula, University of Exeter Medical School, University of Exeter, Exeter, Devon, UK
- ³ Health Economics at Bristol, Population Health Sciences, Bristol Medical School, University of Bristol, Bristol, UK
- ⁴ Centre for Academic Child Health, Bristol Medical School, University of Bristol, Bristol, UK
- ⁵ Patient and Public Involvement Group (PenPIG), University of Exeter Medical School, University of Exeter, Exeter, Devon, UK
- ⁶ Department of Health and Social Sciences, University of the West of England, Bristol, UK

1 Background

Public and patient involvement (PPI) in research is “research being carried out ‘with’ or ‘by’ members of the public rather than ‘to’, ‘about’ or ‘for’ them” [1]. The role and presence of PPI in health research is developing internationally [2–5]. PPI has particularly evolved over the last 2 decades in the United Kingdom (UK), to the point that it is now embedded in the policies of the National Health Service (NHS) [6] and the culture of health services research [7]. However, to date PPI has not been so explicitly prominent in the context of health economics (HE) research. This is beginning to change, and focussed discussion of PPI in HE research is growing [8–10].

There is a broad body of literature on PPI practices, frameworks, and standards. For example, “UK Standards for Public Involvement” was recently released to encourage “better public involvement for health and social care research”. This sets out hallmarks of good public

Key Points for Decision Makers

There are five main principles which may help health economists consider and approach patient involvement: Planning, Approach selection, Continuous involvement, Team building, and Sensitivity.

A “task-based” approach to patient involvement can help with the more technical aspects of health economics research.

The development of ongoing patient–health economist partnerships can enrich patient involvement, ensuring it is non-tokenistic, meaningful, and helpful.

involvement such as flexibility, sharing and learning, and respect for each other [11]. Such approaches are key for guiding supportive, non-tokenistic, meaningful, and useful PPI, but there has been little consideration of the particularities of HE research and “what works” in terms of PPI in this specific discipline [12].

Over the last 7 years, we have worked with people with multiple sclerosis (MS) to inform the development of a variety of HE research studies. In this paper, we draw on our experiences, the experiences of those with whom we have collaborated, and our shared perspectives about what could have worked better and what has worked well. We outline the lessons we have learned about involving patients in HE research, and offer some guidance to health economists and patients wishing to undertake research together. We do not seek to add another PPI framework to a crowded “marketplace”, rather we aim to provide a way of thinking for HE researchers who are getting started with PPI, introducing some principles to consider. We hope these reflections will be relevant to a wide range of conditions and aspects of HE research, and complement more prescriptive frameworks.

2 Our Experiences with Patient Involvement in Health Economics

The Health Economics and MS (HEMS) patient involvement group was initially established to inform a specific aspect of a study that aimed to develop a preference-based measure of health-related quality of life for use with people with MS [13]. This involved selecting a subset of the health states described by the measure for inclusion in a preference elicitation survey. We wanted to avoid selecting health states that seemed implausible to respondents, and considered that people with MS were best placed to identify which health states were implausible. We worked with three people with

MS and developed a task to facilitate the identification of implausible health states. This is described in detail elsewhere [9]. Following a debriefing session, during which the researchers and people with MS reflected on their experiences of the task, we agreed on a number of lessons for the researchers to take forward into future patient involvement.

With these lessons in mind, our subsequent work with the HEMS group has been better planned and more inclusive. The group has expanded substantially, through recruitment of additional members from the South West region. Invitations to join the PPI group were placed in local newspapers and MS centres. Invitations were sent to existing PPI groups, such as the Public Involvement Group at the National Institute for Health Research’s (NIHR’s) Applied Research Collaboration South West Peninsula (PenARC) and local MS support groups. There are now 20 members who work with us, as and when their MS permits. Particular attention has been paid to addressing gaps in the membership of the group, particularly people aged under 50, who are in employment, and/or have relapsing–remitting MS [14]. The role of group members is set out in a document based on the INVOLVE role description template [15], with amendments based on input from the HEMS PPI group and the researchers.

The HEMS group has worked with us to develop successful funding applications, with plans for patient involvement integrated into all stages of our research protocols. We meet regularly to review progress and for the group to advise on specific aspects of the research. These have included identifying an appropriate research area and developing the associated research questions for a study; helping to design an interview schedule for cognitive interviews; advising on the content and wording of documentation for research participants; informing the development of resource use questionnaires for economic evaluations; developing attributes for a discrete choice experiment; identifying health-related events that may affect the wellbeing of people with MS; and assisting with the interpretation of results across a range of studies. Their involvement has ensured our research reflects issues of importance to people with MS, improved the suitability of research materials for intended participants (potentially enhancing response rates and data quality), and given people with MS a voice in the interpretation of data.

3 Lessons Learned

These lessons have been developed by the four people who have been involved throughout the lifetime of the HEMS group: the longest serving HEMS member (LT), two health economists (AH and EG), and a PPI practitioner (KB), and have been informed by formal and informal feedback from the wider HEMS group membership. This includes a recent

online video meeting with eight members of the group specifically to discuss these principles. Where possible, we have drawn on available literature to compare our experiences with those of other research groups.

4 Planning

It is considered best practice to build involvement into the overall research plan at the application stage; indeed, many funders require this [16]. We have found that making involvement activities an integral part of the research from the outset has enabled us to identify which areas of a study require focused involvement activities and to formulate plans for monitoring and oversight of the research programme. When developing research ideas, we have found it useful to ask ourselves, “What information do we need for this research study that we can only (or best) get from patients?” A clear aim regarding the intended outputs from any involvement activity, and how these will be used to guide the research, is essential [10]. Previous research has found that involving people early in the research process, and setting clear goals and plans for involvement, engenders a sense of “ownership” of the research and enhances the impact of involvement [17]. It also provides a basis from which to determine which involvement methods might be appropriate to meet the aims of each aspect of the planned involvement [10, 18].

A crucial aspect when planning involvement activities is to ensure that the background and purpose can be communicated coherently to those involved in the research [10]. One barrier to involving patients in HE research that has been identified is that many of its main concepts and research methods are highly technical and can be inaccessible to lay audiences [12]. We have found the key is to go back to the basics of what we are trying to achieve and to clearly define any core concepts, e.g. “plausibility”. A particularly useful piece of advice from our PPI practitioner is that patient involvement materials do not need to be technically correct in the way that would be expected by an academic audience. Rather, they should convey sufficient information to enable patients to understand the purpose of the research and to engage with it meaningfully. For example, using the description “an experiment where you are asked to make choices” might be a useful route in to discussing discrete choice experiments.

In our experience, and as others have found [18], designing and planning involvement activities requires investment of time and resources, to ensure they generate intended outputs and make sense to the patients involved. We anticipate that designing involvement activities will become easier as more papers of this type are published, building a literature

base that provides sources of different approaches for involvement in HE research.

Plans for involvement also need to be responsive to changing circumstances. An obvious example is the impact of the coronavirus disease 2019 (COVID-19) pandemic. There may be major changes in terms of treatments, etc., that arise unexpectedly and change the landscape for patients. Risks and mitigations in relation to PPI can be considered alongside other aspects of HE research, in order to maximise its flexibility to altered conditions.

5 Approach Selection

Patients should be involved in a meaningful, non-tokenistic way that will provide the required inputs and insights [18, 19]. In our view, there are two broad approaches to involvement: task-based and discussion-based approaches. Each offers their own advantages and disadvantages, and will be more or less appropriate depending on the purpose and nature of any given research activity. This corresponds to Dudley et al’s. [17] distinction between “focused” and “diffuse” impacts of PPI in relation to clinical trials, where the former represent specific effects on particular aspects of a study and the latter represent more general, less tangible benefits arising from researchers and PPI contributors working together.

In our research, discussion-based approaches have worked well for generating “diffuse” impacts and revealing “unknown unknowns”. These have included developing research questions, contributing to funding applications, overseeing research programmes, assisting with planning, and advising on communication with lay audiences. Discussion-based approaches facilitate the identification of broad themes of importance to patients, and can provide crucial information that would otherwise be inaccessible to researchers who lack lived experience of a condition.

Where the research activity is more directed towards shedding light on “known unknowns”, we have found that a task-based approach provides a useful framework for guiding detailed work on highly specific aspects of a study to generate “focused” impacts. The identification of health-related events that affect the wellbeing of people with MS and of attributes for a discrete choice experiment are two examples of “known unknowns” that were addressed successfully via a task-based activity. More detail on these is provided in the “Appendix” (see the electronic supplementary material). The HEMS group have often commented on their enjoyment of using task-based approaches, which they have described as “hard work”, but “thought-provoking” and “fascinating”. The direct relationship between the tasks and the intended impacts on the research has ensured that exercises “did not feel tokenistic” and gave them “a sense of achievement”.

Task-based approaches mandate a narrow focus, which is beneficial for developing aspects of a study where broader opinions, insights, and experiences would not impact the research design. This is advantageous in HE, where many research techniques are highly prescriptive, with fixed parameters that are not open to influence from patient involvement (e.g. preference elicitation techniques, discrete choice experiment designs) [10, 12]. In such cases, the use of a discussion-based approach could be considered disingenuous, as the broader inputs this generates could not be used to influence the research design without compromising the integrity of the research from an economic perspective [12], and ineffective, as it may not produce the precise information required. Nonetheless, the focused nature of a task-based approach can result in issues of importance to patients being missed, and provides little opportunity to challenge orthodox HE research practice from a patient perspective. Thus it is important to acknowledge that this is not an “either-or” dichotomy, and that the two approaches can be used in concert. We have found that dividing up a session into discussion-based and task-based segments provides a useful balance of specific, focused information and broader insights.

6 Continuous Involvement

The most important lesson that the researchers took from their initial foray into patient involvement [9] was the need to involve patients in developing patient involvement. In subsequent feedback, the HEMS group pointed out that various problems encountered during this session could have been avoided if we had worked with them to design the task. This highlights the importance of involving patients meaningfully at all stages of the research process [16].

In our subsequent research, the continuous involvement of the HEMS group has both improved our research by integrating a patient perspective throughout the duration of each study and provided the HEMS group with a context for any specific involvement activities. This, coupled with the involvement of HEMS group members in the design of involvement activities, has resulted in clearer, more accessible activities that run smoothly, produce the intended (and some useful unintended) outputs and are more enjoyable for everyone involved. We have also found it invaluable to include funding for an experienced PPI practitioner as part of the research team, to provide specialist support for this work throughout the project. The continuity of involvement by the HEMS group has extended over a number of research programmes, enabling both group members and researchers to engage in a continuous process of gaining expertise and insight, which they then apply to future work [10, 19].

We acknowledge that this scale and duration of involvement places a demand on people who have other things to do with their time, and are also dealing with a long-term condition. This can make involvement difficult to sustain over time [20]. As recommended [14], we make it clear that HEMS group members can dip in and out over time, depending on their health or other circumstances, without need for explanation. We make use of various modes of involvement, including group meetings, one-to-one conversations, email, and social media, enabling meaningful involvement without the requirement to attend or speak at meetings [21]. Rather than meeting at fixed time intervals, we schedule HEMS group meetings to coincide with the points in the research programme when their specialist input is required, in order to make best use of their time [17]. It is imperative that patients have the opportunity to receive summaries of research findings and their impacts. These should be conveyed regardless of the length of time since the research was conducted. Better still is the involvement of patients in interpreting HE research findings, considering their implications, and disseminating results.

7 Team Building

When reflecting on our first patient involvement task, the HEMS group felt it would have been beneficial to hold an introductory meeting prior to undertaking the task to build a working relationship and to discuss the broader context of the research. As other researchers have found, taking the time to establish strong working relationships, typified by shared understanding and trust, can generate greater positive impacts from involvement [17, 20, 22]. Successful patient involvement groups tend to feel increasingly safe within their own space over time, enriching and strengthening patients’ input [19].

We have found that the continuous involvement of the HEMS group across a number of projects over the years has enabled us to get to know one another and has engendered a shared sense of ownership of the research programme. In this way, we have become a team, each of us with our own expertise and knowledge to bring to the research. Maintaining, extending, and strengthening these relationships over time has required us all to be flexible in our approach as we are challenged by each other’s perspectives [19]. Crucially, we have aimed to make our involvement sessions enjoyable, to be clear about what impact the HEMS group’s input will have on the research, and to ensure that they know how much we value this input [14]. This has enabled us to increase the number of people with MS involved, and the extent and variety of their involvement.

A realist evaluation study of PPI in health research [20] found that providing informal opportunities for researchers

and PPI contributors to socialise together can help foster good working relationships, and our experience supports this. Scheduling social time before and after each formal HEMS meeting allows everyone to physically and mentally arrive, have an initial catch-up, and introduce any agenda items they wish to raise at the meeting, and provides a shared opportunity to reflect and to consider other issues that the session may have brought up. The group frequently use this as an opportunity to share information, e.g. which is the best company locally for building an outside ramp, and how to access physiotherapy services. It also provides the health economists with a greater awareness of what is important to people with MS, and of how their work could be made more inclusive, accessible, and relevant to patients.

8 Sensitivity

Pandya-Wood et al. [14] have suggested that involvement can be emotionally challenging for patients if it causes them to reflect on negative aspects of their condition. Other research studies have identified barriers to involvement in HE research such as the perception of HE as a highly technical discipline, far removed from real patient experience [10, 12], and controversies around the role of the National Institute for Health and Care Excellence (NICE) in approving new drugs and treatments for use by the NHS [21]. These factors caused us concern regarding our initial involvement activity with the HEMS group, in which we described some quite severe health states that the group members might imagine they would experience in the future.

These concerns proved unfounded. The HEMS group have addressed these issues and other aspects of this complex discipline. There is a need to be considerate and sensitive when working with patients [14], but, as our HEMS co-author remarked, “We come as a hardy bunch with armour already inbuilt!” The development of working relationships over time, described above, and the adoption of an honest and open approach are particularly important here.

9 Conclusion

Our aim in this paper has been to share the PACTS principles: planning, approach selection, continuous involvement, team building, and sensitivity. We have aimed to provide practical guidance and illustrative examples for health economists and patients who wish to work together. There is increased recognition that patients are experts at providing a “lived experience” perspective in HE research, in the same way that clinicians are experts at providing an overview of conditions and HEs are experts in the methodology of their discipline. We hope the PACTS Principles complement

existing PPI approaches and frameworks and provide useful foundations for health economists when considering patient involvement, and will ultimately build towards the development of practical guidance for patient involvement in HE research.

Acknowledgements The authors wish to acknowledge the Health Economics and Multiple Sclerosis (HEMS) PPI Group and the Plymouth & Tamar Valley Branch of the MS Society Young Persons Group.

Author Contributions All listed authors (1) made substantial contributions to the conception or design of the work or the acquisition, analysis, or interpretation of data; (2) drafted the work or revised it critically for important intellectual content; (3) approved the version to be published; and (4) agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Declarations

Funding This work was funded by the MS Society and supported by the National Institute for Health Research (NIHR) Collaboration for Leadership in Applied Health Research and Care South West Peninsula. The views expressed are those of the authors and not necessarily those of the NHS, the NIHR, or the Department of Health.

Conflicts of Interest Not applicable.

Availability of Data and Material Not applicable.

Code Availability Not applicable.

References

1. INVOLVE. National Institute for Health Research, London. 2020. <https://www.invo.org.uk/find-out-more/what-is-public-involvement-in-research-2/>. Accessed 19 Feb 2020.
2. National Health and Medical Research Council. Statement on consumer and community involvement in health and medical research. Australia: Consumers Health Forum of Australia; 2016.
3. Canadian Institutes of Health Research. CIHR’s Framework for Citizen Engagement. Partnerships and Citizen Engagement Branch. Ottawa, Canada: Canadian Institutes of Health Research; 2012.
4. Minister of Science and Innovation and Minister of Health. New Zealand Health Research Strategy: Public discussion document. Wellington, New Zealand: Ministry of Health; 2016.
5. National Institutes of Health. Public Involvement at National Institute of Mental Health. 2020. <https://www.nimh.nih.gov/outreach/public-involvement/index.shtml>. Accessed 19 Feb 2020.
6. Department of Health. NHS Constitution. London: HMSO; 2015.
7. INVOLVE. National Institute for Health Research. 2020. <https://www.invo.org.uk/>. Accessed 19 Feb 2020.
8. van Voorn G, Vemer P, Hamerlijnck D, Ramos I, Teunissen G, Al M, et al. The Missing Stakeholder Group: why patients should be involved in health economic modelling. *Appl Health Econ Health Policy*. 2016;14:129–33.
9. Goodwin E, Boddy K, Tatnell L, Hawton A. Involving members of the public in health economics research: insights from selecting health states for valuation to estimate quality-adjusted

- life-year (QALY) weights. *Appl Health Econ Health Policy*. 2018;16(2):187–94.
10. Kandiyali R, Hawton A, Cabral C, Mytton J, Shilling V, Morris C, et al. Working with patients and members of the public: informing health economics in Child Health Research. *Pharmacoeconomics Open*. 2019;3(2):133–41.
 11. UK Public Involvement Standards Development Partnership group. UK standards for public involvement: Better public involvement for better health and social care research. 2019. <https://drive.google.com/file/d/1U-IJNJCfFepaAOruEhzz1TdLvAcHTt2Q/view>. Accessed 12 Feb 2020.
 12. O’Shea E, et al. Knowledge of public patient involvement among health economists in Ireland: a baseline audit. *HRB Open Res*. 2019;2:4. <https://doi.org/10.12688/hrbopenres.12896.1>.
 13. Goodwin E, Green C, Spencer A. Estimating a preference-based index for an eight dimensional health state classification system derived from the Multiple Sclerosis Impact Scale (MSIS-29). *Value Health*. 2015;18:1025–36.
 14. Pandya-Wood R, Barron DS, Elliott J. A framework for public involvement at the design stage of NHS health and social care research: time to develop ethically conscious standards. *Res Involv Engagem*. 2017;3:6. <https://doi.org/10.1186/s40900-017-0058-y>.
 15. INVOLVE. Briefing note five: Be clear with the people you want to involve. Template two: role description. National Institute for Health Research, London. 2020. Accessed 23 Jun 2020.
 16. NHS Health Research Authority. Public Involvement. 2020. <https://www.hra.nhs.uk/planning-and-improving-research/best-practice/public-involvement/>. Accessed 23 Jun 2020.
 17. Dudley L, Gamble CG, Preston J, Buck D, The EPIC Patient Advisory Group, Hanley B, Williamson P, Young B. What difference does patient and public involvement make and what are its pathways to impact? Qualitative study of patients and researchers from a cohort of randomised clinical trials. *PLoS One*. 2015;10(6):e0128817. <https://doi.org/10.1371/journal.pone.0128817>
 18. Pizzo E, Doyle C, Matthews R, Barlow J. Patient and public involvement: how much do we spend and what are the benefits? *Health Expect*. 2015;18(6):1918–26.
 19. Mullins CD, Abdulhalim AM, Lavalley DC. Continuous patient engagement in comparative effectiveness research. *JAMA*. 2012;307:1587–8.
 20. Wilson P, Mathie E, Keenan J, McNeilly E, Goodman C, Howe A, Poland F, Staniszeska S, Kendell S, Munday D, Cowe M, Peckham S. ReseArch with Patient and Public involvement: a RealisT evaluation – the RAPPORT study. *Health Serv Deliv Res*. 2015;3(38)
 21. Gibson A, Welsman J, Britten N. Evaluating patient and public involvement in health research: from theoretical model to practical workshop. *Health Expect*. 2017;20(5):826–35.
 22. Crocker J, et al. Is it worth it? Patient and public views on the impact of their involvement in health research and its assessment: a UK-based qualitative interview study. *Health Expect*. 2017;20(3):519–28.