Patient and public involvement in cancer-associated thrombosis research: necessary or glorified tokenism?

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ABSTRACT

The advantages of patient and public involvement (PPI) in research are becoming more widely known, however different research organizations have different rates of adoption. Comparably, some groups provide tokenistic participation in the research process, which is inconsistent with the extent to which PPI partners are truly involved. Recent developments in the field of cancer-associated thrombosis (CAT) research have shown how crucial PPI is to the foundation of the entire research process, from formulating the research question to disseminating the findings. This manuscript aims to present an overview of PPI within the framework of CAT research and demonstrate how, when used appropriately, PPI can improve a project's overall success, rigor, and relevance.

Introduction

There is an Apocryphal story about the great Louis Armstrong who, when asked the question "What is jazz?" replied with the often misquoted answer:[1] "Man! If you gotta ask, you'll never know!"

Such a response comes to mind when I am asked by academic colleagues to explain the point of public involvement and public engagement in biomedical research. The question usually arises following a discussion in which I have challenged their assertion that having a layperson cast their eyes over a patient information sheet for a research study constitutes sufficient public involvement and public engagement. Recent years have seen an increased interest in patient and public involvement in research such that many research groups will have representation in their trial management groups. However, the contribution of public involvement representatives is variable, particularly when researchers have considered their activities to be a "box-ticking exercise" rendering their involvement tokenistic.[2,3] This paper shall discuss the merits of embracing public involvement within our research and shall include suggestions on how to optimize such activities. Whilst it is readily acknowledged that many examples of excellent public involvement exist around the globe, for the purposes of this paper, the focus shall be on undertakings within the United Kingdom (UK). It shall also give examples of where public involvement has been used successfully in cancer-associated thrombosis (CAT) research.

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Definitions

One of the biggest challenges in embedding meaningful public involvement in research is to inform people what it is in a way that the research community can understand, what the benefits are and how to embed it in such a way that it makes a real difference rather than just look good. In order to implement a new development, one must first understand what new concept or activity you are trying to introduce. For the sake of this paper definitions of public involvement and public engagement shall be as follows:

Public involvement (PI) in research is defined by the UK Health Research Authority as "research carried out 'with' or 'by' members of the public, rather than 'to', 'about' or 'for' them. It means that patients or other people with relevant experience contribute to how research is designed, conducted and disseminated. It does not refer to research participants taking part in a study".[4]

Public engagement (PE) as defined by the UK National Coordinating Centre for Public Engagement describes "the myriad of ways in which the activity and benefits of higher education and research can be shared with the public. Engagement is by definition a two-way process, involving interaction and listening, with the goal of generating mutual benefit".[5]

Confusion often arises when the term PE is erroneously used

as an umbrella term to describe both PE and PI. Many descriptions for public involvement and public engagement exist with some being used interchangeably, inevitably leading to confusion and sub-optimal practice.

Drivers for public involvement

The main overarching driver for PI in research is the fact that it supports the development, conduct and overall success of good research. The renowned theoretical physicist Richard Feynman was considered one of the most influential and inspiring scientists of the 20th century. An alumnus of Robert Oppenheimer's Manhattan Project, Nobel Laureate for his work on quantum physics and member of the Rogers Commission into the Challenger space shuttle disaster, he was also an amazing raconteur with a reputation for making science exciting and interesting. He suggested we should undertake research for "the pleasure of finding things out" a sentiment used as the title of a documentary made about him and a separate book of papers and correspondence collated by his children after his death.[6]

Whilst this is an admirable sentiment and no doubt a sufficient and achievable driver in some specialties, such philosophy lends itself less well to research in clinical medicine. For one, clinical studies are required to satisfy the scrutiny of research ethics committees to ensure there is a cogent case for the research and involvement of patient participants is justified. Funding bodies are unlikely to fund a study just because it sounds interesting, they will want to be convinced that there is not only a need for the research but also there is a strong likelihood of recruiting participants. Furthermore, they will want to know if there is a strong likelihood that the results will impact on clinical practice. These answers are unlikely to be accurately answered without the input and advice from laypersons who will see the rational importance of the study and acceptability of the intervention through a different viewpoint.

Many funders in the UK have patient and public partners on their funding panel and will expect applications to outline the amount of PI in the development of the application and how they plan to involve PI partners in the ongoing project. From the author's experience, some funding requests have been rejected on the grounds of insufficient PI or for not costing it into the bid.

Whilst PI representation is not mandatory when presenting the project at the research ethics committee meeting, it can be very useful if any questions arise regarding the patient perspective or concerns raised by lay members of the committee. Finally, the role of PI is becoming more important when looking to publish the results in peer-reviewed journals. The British Medical Journal amongst others, requires authors to outline how patients were involved in the delivery of the research, with more of their manuscripts including patient perspectives within the narratives.

Benefits of public involvement

Rather than undertaking PI in research because funding bodies, ethics committees and journals say we should, it is worth considering that rather than being the "new fashion", it is also a means by which we can undertake better research. The UK Health Research Authority suggests that academic teams engaged in PI conduct better research and undertake better studies because: i) they design studies that are of greater relevance to participants; ii) studies are more likely to be acceptable to participants; iii) participants find the study information to be more understandable; iv) the patient experience of research is better and provide a better experience of research; v) study results are communicated better to participants at the end of the study.[4]

Conducting research with public involvement

Undertaking meaningful involvement of patients and the public in health and social care research should follow four agreed principles which are: involve the right people, involve enough people, involve those people enough, and describe how it helps. These are discussed in the following paragraphs.

Principle 1: involve the right people

This means you should be involving people with lived experience of the condition being studied. Sometimes this may not be possible for various reasons; when studying patients with incurable illnesses or who are too unwell to contribute, it may be better to involve carers or significant others who have some experience and understanding. Sometimes representation from patient groups may be possible especially if they are able to act as a conduit between the researchers and a wider public population. In addition to having lived experience of the condition, it is also important to ensure the PI contributor is representative of the population associated with the condition. For example, some conditions may be associated with lifestyle choices or socioeconomic deprivation. Others might target patients of a particular gender, age, ethnicity or geographical region. It does not make sense, therefore, to only have a pool of PI contributors consisting solely of white middle-class retired gentlemen who divide their time between golf and meetings at the Rotary Club.

Principle 2: involve enough people

In order to understand the breadth of views on issues of importance to the target recruitment population, there is a need for sufficient PI representation to gain an accurate perspective of the different people whom the research is intended to benefit. Numbers will vary according to the scope of the study but a single contributor will rarely be able to covey the views and needs of the whole study population. Most of the CAT studies we have undertaken will have a minimum of two PI contributors, from different backgrounds and experiences.

It is also worth considering different roles for different contributors; not everyone needs to be a member of the project team. Some may wish to focus on reviewing the recruitment pathway or patent information literature. Others may review the acceptability of planned assessment tools or even the form of how the assessment will be conducted.

Principle 3: involve those people enough

PI contributors should be given the opportunity to be involved in as many aspects of the research project as possible. Ideally, they should be involved at the planning stage, before

funding has been awarded. It will enhance the planning of the study and ensure its relevance to the patient population. It may also identify potential pitfalls to recruitment. Examples of activities that contributors might undertake are listed in Figure 1.

Principle 4: describe how it helps

There is an expectation that researchers inform funders and regulatory authorities including the REC to describe: i) those involved in the study as PI contributors including the relevant experience they brought to the project; ii) what activities PI contributors undertook; iii) how their involvement benefit the study, *i.e.*, in what way they helped the study become more relevant, acceptable to study participation; iv) how study results are to be shared with study participants (if they wish to know) and other stakeholders.

Evaluating public involvement contribution

The UK Public Involvement Standards Development Partnership developed a set of standards against which researchers could benchmark their activity.[7] These are outlined in Figure 2. Within CAT research, the UK standards were used to evaluate PI during the Hospice Inpatient Deep vein thrombosis Detection study (HIDDen), a multicentre, prospective, longitudinal, observational study to explore the prevalence, symptom burden and natural history of venous thromboembolism in people with advanced cancer.[8,9] This was led by the study PI contributor lead who had also been involved in the development of UK standards. They concluded that all six standards were met with the greatest opportunities in 'working together' and 'support and learning'. Meeting the 'governance' standard was less complete; with evidence of participation in decision-making but little involvement in management, regulation, and leadership. The experience of benchmarking PI activity against the UK standards revealed that such appraisal was largely subjective and ideally PI involvement should be evaluated in real time so that involvement can be proactively managed. Recently, the Marie Curie Research Group and Wales Cancer Research Centre at Cardiff University have developed the Public Involvement in Research Impact Toolkit (PIRIT).[10] This is free to use and available online (https://www.cardiff.ac.uk/marie-curie-research-centre/patient-and-public-involvement/public-involvement-in-research-impact-toolkit-pirit). It was designed as a set of pragmatic tools to support researchers working with public contributors who aim to: i) proactively integrate PI into the research project through planning; ii) allow teams to track the activity of PI public contributors and evaluate the difference they have made to the research; iii) allow teams to report this in a format that benchmarks activities against the UK standards for PI.

These consist of the PIRIT planning tool and the PIRIT tracking tool. The PIRIT planning tool is a checklist of possible PI activities that may be available and follows the research pathway allowing teams to objectively measure if and how they meet the relevant standards. The PIRIT tracking tool comprises a spreadsheet to record when and how the public contributed to the research. More specifically, it will record what the activities hoped to achieve, whether their involvement made any difference, why this mattered and how this relates to the standards.

Public involvement in cancer-associated thrombosis research

The role of PI in the HIDDen study has already been discussed.[9] However, following its publication, PI in the dissemination and reflection stage of the research further influenced the next research project. With the support of the lead PI contributor, a round table discussion was organized with representation from all relevant UK professional and patient organizations. The data were presented and discussed, with particular emphasis on how the research would influence practice and whether there were ongoing unanswered questions to answer. Through this forum, the patient organization representatives gave a very strong steer on what questions were important to them and this formed the basis of the follow-up study HIDDEN2.[11]

Currently, the SERENITY study is underway; this is an ambitious 7-phased multicenter European mixed methods research program that aims to develop and subsequently evaluate a shared decision-making tool for the continuation or deprescribing of

Before funding awarded
- Developing and prioritizing research questions;
- Identifying outcome measures of relevance to the population being studied;
- Contributing to research methods with respect to practicalities such as how to optimize recruitment, minimize participant burden, identify potential pitfalls and obstacles;
- Contributing to grant application.

Once funding awarded
- Commenting on full protocol;
- Contribute to Research Ethics Committee (REC) applications;
- Attending REC meeting to discuss acceptability of study to participants;
- Co-designing and commenting on participant information sheets and other patient facing documents.

During the research
- Active participation on research advisory groups and steering groups;
- Supporting aspects of the research e.g. co-facilitating focus groups, questionnaires or distributing questionnaires.

After the research completed
- Contributing to dissemination.

Figure 1. Examples of activities undertaken by public involvement contributors.

- Inclusive opportunities:
  - Offer public involvement opportunities that are accessible and that reach people and groups according to research needs;
- Working together:
  - Work together in a way that values all contributions, and that builds and sustains mutually respectful and productive relationships;
- Support and learning:
  - Offer and promote support and learning opportunities that build confidence and skills for public involvement in research;
- Communications:
  - Use plain language for well-timed and relevant communications, as part of involvement plans and activities;
- Impact:
  - Seek improvement by identifying and sharing the difference that public involvement makes to research;
- Governance:
  - Involve the public in research management, regulation, leadership and decision making.

Figure 2. United Kingdom public involvement standards for research.[7]

antithrombotic medicines in patients with advanced cancer near the end of life.[12] Public involvement has been embedded in the project with each phase having PI activity planned. The study is being conducted across fourteen different academic institutions in eight European countries, which have differing levels of knowledge, experience and confidence in PI activity. It has thus been a dynamic, iterative educating experience for many researchers. One very apparent thing, however, is the consensus that the PI partners and the PI coordinator are an integral and essential component of the study group. Any thoughts of tokenism have long dissipated.

Conclusions

For many, public involvement is one more of a long line of tokenistic activities that do little other than symbolize academic institutions acceding to public pressure. They see it as a necessary hurdle to jump over in order to get a study funded, ethically approved and, ultimately, published in a high-profile journal. Such attitudes do result in tokenism being practiced within their own particular research groups. However, organizations that embrace the public as true partners and advisers, derive the benefit of their involvement very early on and reap the rewards of better-designed, successfully recruited-to studies of true relevance to the population being studied.

Thinking back to the original quote in this paper regarding Louis Armstrong's response is, in fact, an urban myth: as accurate as quotes such as "Play it again Sam", or "Luke, I am your father."

The true response by Armstrong, and on reflection, far more apt when considering it in the context of defining public involvement, is: "If you still have to ask, shame on you".[1]

References

1. Jones M, Chilton J, Feather L. Salute to Satchmo. London: I.P.C. Specialist & Professional Press Ltd. 1970:155.
2. Andrews LM, Allen H, Sheppard ZA, et al. More than just ticking a box… how patient and public involvement improved the research design and funding application for a project to evaluate a cycling intervention for hip osteoarthritis. Res Involv Engagem 2015;1:13.
3. Supple D, Roberts A, Hudson V, et al. From tokenism to meaningful engagement: best practices in patient involvement in an EU project. Res Involv Engagem 2015;1:5.
4. Health Research Authority (HRA). Public Involvement. 2022. Available from: https://www.hra.nhs.uk/planning-and-improving-research/best-practice/public-involvement
5. Engagement NC-oCfP. Introducing Public Engagement. 2023. Available from: https://www.publicengagement.ac.uk/about-engagement/what-public-engagement
6. Feynman C, Feynman M. The Pleasure of Finding Things Out. 1st ed. United States: Perseus Books 1999:270.
7. Partnership UPISD. UK Standards for Public Involvement: Better public involvement for better health and social care research. NIHR 2019. Available from: https://sites.google.com/nihr.ac.uk/pi-standards/home
8. White C, Noble S, Swan F, et al. 'Hospice inpatient deep vein thrombosis detection (HIDDen) in advanced non-malignant diseases': a longitudinal pilot study. BMJ Support Palliat Care 2022;12:e767-e70.
9. Seddon K, Elliott J, Johnson M, et al. Using the United Kingdom standards for public involvement to evaluate the impact of public involvement in a multinational clinical study. Res Involv Engagem 2021;7:22.
10. Centre. MCRCatWCR. Public Involvement in Research Impact Toolkit (PIRIT) https://www.cardiff.ac.uk/marie-curie-research-centre/patient-and-public-involvement/public-involvement-in-research-impact-toolkit-pirit2023
11. Kitson T, Osborne E, Noble S, et al. HIDDEN2: Study protocol for the hospital deep vein thrombosis detection study in patients with cancer receiving palliative care. BMJ Open 2023;13:e073049.
12. Goedegebuur J, Abbel D, Accassat S, et al. Towards optimal use of antithrombotic therapy of people with cancer at the end of life: A research protocol for the development and implementation of the SERENITY shared decision support tool. Thromb Res 2023;228:54-60.