OPINION ARTICLE

How to incorporate patient and public perspectives into the design and conduct of research [version 1; referees: 3 approved, 2 approved with reservations]

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Abstract

International government guidance recommends patient and public involvement (PPI) to improve the relevance and quality of research. PPI is defined as research being carried out 'with' or 'by' patients and members of the public rather than 'to', 'about' or 'for' them (http://www.invo.org.uk/). Patient involvement is different from collecting data from patients as participants. Ethical considerations also differ. PPI is about patients actively contributing through discussion to decisions about research design, acceptability, relevance, conduct and governance from study conception to dissemination. Occasionally patients lead or do research. The research methods of PPI range from informal discussions to partnership research approaches such as action research, co-production and co-learning.
This article discusses how researchers can involve patients when they are applying for research funding and considers some opportunities and pitfalls. It reviews research funder requirements, draws on the literature and our collective experiences as clinicians, patients, academics and members of UK funding panels.

Keywords

Public and Patient Involvement, Public Engagement, Qualitative research, Research Methods, Co-production, Partnership approaches

Open Peer Review

Referee Status: ✓ ? ✓ ? ✓

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Introduction

Patient and public involvement (PPI) is recommended from the earliest research stages through to dissemination of the findings[1–6]. In the UK, INVOLVE[3] states that research should be done with and by patients, but what does this mean for researchers and patient partners when starting a study? International resources are available (Box 1) and six UK PPI standards are being tested to see if they work in practice[7]. Table 1 summarises on-line guidance for research applications to international government funding programmes that endorse involving patients and the public. Language varies internationally and is evolving as patients take a more central role in deciding what research is done and how. Box 2 provides some definitions which are derived from the INVOLVE jargon buster[8] and international resources (Table 1). PPI includes patients, potential patients, families, carers, patient groups and members of the public who use or have access to health and social care services[3]. We refer to this broad group as 'patients' to distinguish them from clinicians and academics. This is consistent with Canadian guidance, which defines 'patients' as 'an overarching term inclusive of individuals with personal experience of a health issue and informal caregivers, including family and friends'[9]. However, 'patients' may include people who do not describe themselves in this way. People may self-care for their condition and general public contributions can add value to research questions. Other relevant international terms, for example stakeholder involvement, consumer involvement, knowledge user engagement and patient orientated research are described in Supplementary File 1, Section A.

PPI is put into practice through patients discussing, helping to make decisions and occasionally doing research in order to enhance study relevance, design, conduct and governance. There is no 'one-size-fits-all' approach. Flexibility is required to tailor patient involvement to the topic, research question, methods and resources available. This article describes steps that researchers and patient partners can follow when preparing a research funding application (Box 3). We refer throughout the article to an illustrative example of a researcher who wants to do a study to improve outcomes for patients with migraine, and we provide examples from the literature and authors' experiences.

Steps for how to involve patients and the public when applying for research funding

Understand what patient and public involvement is

At the outset, it is important to understand the theory underpinning PPI. In depth reviews and discussion of theory are available[1,4,10–13] and suggest that, depending on the circumstances, PPI will:

- ensure that the research questions and outcomes really matter to patients
- provide perspectives that complement or challenge those of researchers and clinicians
- make research more relevant to the people whom it is designed to benefit
- ensure that proposed research will be acceptable to patients so that they will be willing to participate
- improve the quality of research
- offer lay knowledge that is either independent for the purpose of governance, or specific to the focus of study to enhance its design or conduct
- make research more equitable and ethical, particularly when publicly funded
- improve dissemination to reach wider lay audiences
- increase the likelihood that research will be implemented into everyday practice and impact on patient care
- enable patients to feel that their voice matters.

All of the above could reduce research waste[14–16] if PPI is put into practice in ways that ensure that research is meaningful, acceptable, ethical and useful.

How does patient involvement differ from patient participation in research?

Patient perspectives can be sought through patient *involvement* and through patients *participating* in surveys, interviews or focus groups to provide data for others to analyse, interpret and act on. The authors have observed that in grant applications and study protocols, PPI is often conflated with qualitative research or patient opinion surveys. Collecting data from patients can be important to gain diverse or representative views, but it is different from PPI and both are often needed (Table 2). Discussion with patients at a workshop can seem similar to collecting data in a focus group, because both involve listening to patients' perspectives, but the context and outcomes from listening differ. PPI means that researchers are in a continuing and reciprocal relationship with patients and make decisions with them about the research. In qualitative research, researchers listen to patients in order to

Box 1. Useful resources for patient and public involvement (listed alphabetically)

International endorsement of public and patient involvement in research

Australian Government National Health and Medical Research Council: Consumer and Community Involvement: https://www.nhmrc.gov.au/research/consumer-and-community-involvement

Australian Government National Health and Medical Research Council: Statement on Consumer and Community Participation in Health and Medical Research (the Statement on Participation): https://www.nhmrc.gov.au/guidelines-publications/r22

Canadian Institutes of Health Research, Strategy for Patient-Oriented Research (SPOR): A coalition dedicated to the integration of research into care: http://www.cihr-irsc.gc.ca/e/41204.html

Cochrane Consumer Network. Statement of Principles for Consumer Involvement in Cochrane: http://consumers.cochrane.org/news/statement-principles-consumer-involvement-cochrane

Cochrane Training. Involving People learning resource relating to systematic reviews, developed by the ACTIVE (Authors and Consumers Together Impacting on eVidencE) project: http://training.cochrane.org/ACTIVE

European Patient Academy (EUPATI) is a network of European National Platforms which supports the integration of patient involvement across the entire process of medicines research and provides training. This includes the pharmaceutical industry and regulatory agencies. https://www.eupati.eu/

European Health 2020 Strategy calls for civil society engagement to improve health: http://www.euro.who.int/en/publications/abstracts/health-2020-a-european-policy-framework-supporting-action-across-government-and-society-for-health-and-well-being

Gordon and Betty Moore Foundation and The American Institutes for Research: A roadmap for patient and family engagement in health and research: http://patientfamilyengagement.org/

Health Technology Assessment International Patient and Citizen Involvement: www.htai.org/interest-groups/patient-and-citizen-involvement/

Nuffield Council on Bioethics (2012) Emerging biotechnologies: technology, choice and the public good: http://nuffieldbioethics.org/project/emerging-biotechnologies/

Ottawa Charter for Health Promotion. World Health Organisation 1986: (http://www.who.int/healthpromotion/conferences/previous/ottawa/en/index1.html)

Patient Centered Outcomes Research Institute (PCORI) standards: https://www.pcori.org/research-results/about-our-research/research-methodology

PCORI Engagement Rubric: https://www.pcori.org/sites/default/files/Engagement-Rubric.pdf

The European Group on Ethics in Science and New Technologies. (2015). Opinion on the ethical implications of new health technologies and citizen participation. Europa: https://ec.europa.eu/research/ege/pdf/opinion-29_ege_executive-summary-recommendations.pdf

US Department of Health and Human Services: Public Involvement with the National Institutes of Health: https://www.nih.gov/about-nih/what-we-do/get-involved-nih/public-involvement-nih

Key UK-based resources and organisations

Healthtalk.org. Patient and public involvement in research: personal stories of patient involvement in research: http://www.healthtalk.org/peoples-experiences/medical-research/patient-and-public-involvement-research/topics

INVOLVE: supports active public involvement in NHS, public and social care research. Funded by NIHR. http://www.invo.org.uk/. There are useful pages on 'Budgeting for Involvement Guidance': http://www.invo.org.uk/wp-content/uploads/2014/11/10002-INVOLVE-Budgeting-Tool-Publication-WEB.pdf and an Involvement Cost Calculator: http://www.invo.org.uk/resource-centre/payment-and-recognition-for-public-involvement/involvement-cost-calculator/

invoDIRECT is a directory of organisations, networks and groups that support active public involvement in research and helps people to identify activity in their area of interest. http://www.invo.org.uk/communities/invodirect/.

invoNET is a network of people who are building the evidence knowledge and learning about public involvement in research: http://www.invo.org.uk/communities/invonet/.

James Lind Alliance: bring patients, carers and clinicians together in Priority Setting Partnerships to identify and prioritise the top uncertainties, or unanswered questions, about the effects of treatments: http://www.jla.nihr.ac.uk/

National Co-ordinating Centre for Public Engagement (NCCPE) has sections for researchers (and others) to explore, support, plan and do public engagement. It runs training courses and helps Universities to engage with the public: https://www.publicengagement.ac.uk/

NHS Health Research Authority: protects and promotes the interests of patients and the public in health and social care research and has top tips on public involvement in grant applications: https://www.hra.nhs.uk/planning-and-improving-research/research-planning/public-involvement/

NICE's approach to public involvement in guidance and standards: a practical guide (2015): https://www.nice.org.uk/media/default/About/NICE-Communities/Public-involvement/Public-involvement-programme/PIP-process-guide-apr-2015.pdf

NIHR Going the Extra Mile strategy: https://www.nihr.ac.uk/patients-and-public/documents/Going-the-Extra-Mile.pdf

NIHR Patient and Public involvement in research. https://www.nihr.ac.uk/patients-and-public/

NIHR Public involvement standards development: a project aiming to improve the quality and consistency of public involvement (PI) in research through the development and introduction of national standards: https://sites.google.com/nihr.ac.uk/pi-standards/home

NIHR Research design service: provides support to health and social care researchers across England on all aspects of developing a grant application including, research design, research methods, identifying funding sources and involving patients and the public. Their advice is confidential and free of charge: https://www.nihr.ac.uk/about-us/how-we-are-managed/our-structure/research/research-design-service/

Patients active in research: A website promoting partnership between patient, carers, members of the public and medical researchers, including case studies of patient involvement in research and opportunities to take part in medical research: https://patientsactiveinresearch.org.uk/

Patients included charters: provide entities with a means of demonstrating their commitment to incorporating the experience and insight of patients into their organisations by ensuring that they are neither excluded, nor exploited: https://patientsincluded.org/

People in research: helps researchers and research organisations to find patients to work with and advertises opportunities for public involvement in NHS, public health and social care research: https://www.peopleinresearch.org/

Research Councils UK Concordat for Engaging the Public with Research: https://www.ukri.org/public-engagement/

Table 1. International government research funding guidance that endorses patient and public involvement (accessed November 2017).

Government general research guidance*	Terminology*	Application form specific guidance	Key features of funding applications
Australian Government National Health and Medical Research Council (NHMRC): Guidelines on Consumer and Community Participation in Health and Medical Research endorse consumers right to make contributions	Consumer and Community Involvement Consumer and Community Participation	NHMRC advice and instructions to applicants: https://www.nhmrc.gov.au/book/nhmrc-advice-and-instructions-applicants-2017/nhmrc-advice-and-instructions-2017 Indigenous populations: https://www.nhmrc.gov.au/book/nhmrc-funding-rules/section-b-project-grants/b-attachment-d-guidance-applicants-address-criteri	Mandatory Plain English Summary. Consumer and community engagement sections are not always mandatory and applicants can decide 'if applicable'. There is either a form section to complete which asks applicants to enter each community engagement activity separately or instructions to upload either a one or two page document. Fellowship applications require information about community engagement and participation for the previous 5 years or 10 years. Some forms have 'if applicable' sections which ask applicants to describe: a) how participants will have access to their own results and how the researchers will be accountable to participants for the overall results of the research: b) how they will ensure consumers are involved in the research and how they will communicate results to participants and the community.
Canadian Institutes of Health Research (CIHR), Strategy for Patient-Oriented Research (SPOR) Framework. Patient, Citizen and Knowledge User Engagement in research should be meaningful throughout the research process, to inform research planning and design.	Patient Orientated Research Patient engagement in research Citizen engagement in research Knowledge user engagement.	Current Opportunities: https://www.researchnet-recherchenet.ca Index of funding related forms: http://www.cihr-irsc.gc.ca/e/797.html Some application forms do not mention patient/citizen engagement, e.g. secondary analysis of existing datasets.	*SPOR funded opportunities* Mandatory research activities relating to the SPOR Framework, e.g. patients engaged as partners; focus on patients' priorities; patients must play a key role in a multi-disciplinary team; a minimum of one Principal Applicant or Principal Knowledge User must be a patient; a workshop, roundtable, public lecture or citizen/patient engagement forum. A patient engagement plan is required Funding: applies a 1:1 matching formula with non-federal government partners. Applicants can request consultant fees for patient engagement experts and costs to facilitate engagement such as compensation, incentives, and the development of orientation and training. *Non SPOR funding opportunities* A lay summary is not always required. Some forms ask applicants to provide a Knowledge/ Technology Users (KTU) plan with activities that will translate the research results outside the academic environment. Funding may be available for indigenous populations. 'Costs related to community mobilisation and engagement, including culturally relevant promotional items such as, tobacco, cloth, and cash reimbursements (in a method acceptable to the individual or community being reimbursed) to compensate community participation; and contracts and/or consultant fees for knowledge translation and communication activities for Elders, community members, and other Knowledge Holders involved in activities related to the Indigenous community'.
European Research Commission. Horizon 2020 "Better Research for Better Health" https://ec.europa.eu/programmes/horizon2020/sites/horizon2020/files/SPH_VisionPaper_02062016.pdf	Involving citizens, end-users and the public sector. Citizen Science. Multi-stakeholder approach. Public engagement. Co-design.	'Science with and for society' is one of five main priorities in the Horizon 2020 Online Manual for applicants: http://ec.europa.eu/research/participants/docs/h2020-funding-guide/index_en.htm Science with and for society http://ec.europa.eu/research/participants/data/ref/h2020/wp/2018-2020/main/h2020-wp1820-swfs_en.pdf	Form requirements vary according to the funding call. Involving patients and citizens is recommended and new approaches are advocated. Citizen science is supported at all stages of research and innovation, with co-design specifically mentioned. Current funding facilitates the establishment of self-sustaining ecosystems for citizen science that will continue beyond the duration of the funds. Guidance encourages institutional changes to sustain new forms of citizen involvement in research and innovation decision making.

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Government general research guidance*	Terminology*	Application form specific guidance	Key features of funding applications
UK Health and Social Care Act 2012 endorses public and patient involvement: http://www.legislation.gov.uk/ukpga/2012/7/contents UK Research Councils Concordat for Engaging the Public with Research.	Public Engagement	The Medical Research Council funds public engagement activities https://www.mrc.ac.uk/research/public-engagement/ 'Pathways to Impact' covers public engagement activities https://www.mrc.ac.uk/funding/guidance-for-applicants/2-the-application/#2.2.5	All Research Council funding applications have an Impact Summary, which is for the public domain. In addition there is a two page attachment called 'Pathways to Impact' where applicants are asked what they will do to make beneficiaries aware of the research and how they will benefit. Impact generating activities are two-way and start when developing the application: https://www.publicengagement.ac.uk/plan-it/funding/public-engagement-and-pathways-impact. Applicants can request project-specific resources and are asked justify costs. Training costs, specialists in public engagement, materials, venue costs and travel expenses are eligible for full economic costing.
UK National Institute of Health Research (NIHR) supports Patient and Public Involvement in research. NIHR Going the Extra Mile strategy. NIHR standards for public involvement are in development.	Public and Patient Involvement (PPI) Co-production.	Application form guidance has a section on PPI: https://www.nihr.ac.uk/funding-and-support/documents/current-funding-opportunities/hta/HTA-Full-Guidance-Notes.pdf Extensive guidance on budgets and a cost calculator are on the INVOLVE website (www.invo.org.uk).	There are mandatory PPI sections and a lay summary in NIHR forms. If applicants are a member of the public, patient /service user or carer, they are asked to provide their knowledge, skills and experience instead of a CV. PPI sections ask: Were patients and the public actively involved in identifying the research topic or prioritising the research questions? Were patients and the public actively involved in preparing this application? Please indicate the ways in which patients and the public will be actively involved in the proposed research. Eligible costs include: payment, rewards, expenses, costs of activities and involvement staffing to support, co-ordinate and facilitate involvement. Payment options include fees, vouchers, donations, gifts, funding for training, honorary appointments. Expenses include travel, subsistence, child care/carer costs, personal assistants, overnight accommodation and home office costs. Involvement activity materials, venues, catering and conference fees can be included, as well as translation, interpretation and support for people with impairments. The form requires full economic costs for PPI and justification of costs. There is no upper limit and external peer review guides funding decisions.
US Department of Health and Human Services: Public Involvement with the National Institutes of Health. Patient Centered Outcomes Research Institute (PCORI) standards.	Patient partnerships Patient-Centered Outcomes Patient engagement Co-learning Reciprocal relationships.	Pre-award user guide: https://www.pcori.org/sites/default/files/PCORI-Online-Pre-Award-User-Guide.pdf PCORI Engagement Rubric for the entire research process includes a financial compensation framework https://www.pcori.org/sites/default/files/Engagement-Rubric.pdf	Researchers partner with patients and other stakeholders from the planning stages through to dissemination of findings to answer patient-centered questions. Focus on outcomes that matter to patients. For some highly technical and methodological projects, patients may not make appropriate partners. A public abstract and an engagement plan covering three stages: planning, conduct and dissemination are required. Applicants are expected to demonstrate how they meet the six PCORI patient engagement principles in their work: reciprocal relationships; co-learning, partnerships, transparency, honesty and trust. Information is required that supports how patient informants and people representative of the population of interest input into decisions about outcomes. Information may come from meetings, surveys or published literature. Applicants are asked to give a detailed budget aligned with engagement activities as outlined in the engagement plan. This includes compensation for patient time and expenses incurred (travel, accommodation, parking, childcare, respite or caregiver expenses. special needs, phone, internet). Applicants are advised not to let cost be a barrier to patient engagement and to include a staff budget to support patient engagement: e.g. recruitment, training, mentoring, co-ordinating and for engagement events.

*Websites for international research guidance, definitions of terms and additional information are provided in Box 1, Box 2 and Supplementary File 1

Box 2. Terminology

Some acronyms for involving people in research

PPI –Patient and Public Involvement: http://www.ukcrc.org/patients-and-public/. In the European Research Commission, PPI means Public Procurement of Innovative Solutions.

PPIE – Public Patient Involvement and Engagement: https://www.nihr.ac.uk/about-us/documents/PPIE-Leadership/NIHR-PPIE-Strategy_2018-19.pdf

PIA – Public Involvement Activities[17]

PCORI – USA Patient-Centered Outcomes Research Institute http://www.pcori.org/program/engagement

NIP – National Involvement Partnership which includes the 4PI – Principles, Purpose, Presence, Process, Impact which are national involvement standards https://www.nsun.org.uk/FAQs/4pi-national-involvement-standards

Definitions

Definitions are derived from the INVOLVE jargon buster[8] and international resources in Table 1 and Box 1.

Participating in research describes people who have consented to provide data for analysis to further knowledge (participants). Historically participants were referred to as 'subjects' of research. 'Participatory research approaches'[18] is used as an umbrella term which covers 'participatory action research'[19,20], co-design[21,22] and co-production of research[23,24]. In our opinion, a more suitable umbrella term is 'partnership approaches'.

Involving. INVOLVE[3] defines public involvement in research as research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them and states that the term 'public' includes:

- Patients, potential patients, carers
- People who use health and social care services
- People from organisations that use services

INVOLVE makes a distinction between the 'public' and people who have a professional role in health and social care.

The European Union (EU) website refers to 'citizen Involvement' which includes upstream priority setting, influencing funding decisions to a more direct downstream involvement of citizens and patients in the use and application of medical knowledge and information. It covers both active citizens who engage from a position of agency as well as those unaware of their contribution[25]. 'Citizen Science' is used as an EU umbrella term which is envisioned as various forms of public engagement with science as a way to promote responsible research and innovation.

Partnership is when people who get actively involved in research have a relationship that involves mutual respect and have an equal voice. This contrasts to someone who is consulted occasionally. PCORI consider that the principle is demonstrated when time and contributions of patients and stakeholder partners are valued and demonstrated in fair financial compensation, as well as in reasonable and thoughtful requests for time commitment. When PCORI studies include priority populations, the research team is committed to diversity across all project activities and demonstrates cultural competency, including people with disabilities, when appropriate.

Reciprocal Relationships is one of six PCORI engagement principles. They are demonstrated when the roles and decision-making authority of all research partners, including patients, are defined collaboratively and clearly stated.

Collaborating is active, on-going involvement in the research process; however, responsibilities are not equally shared like they are in partnerships. Patients may be co-applicants on a grant application, take part in an advisory group or work with researchers to design, undertake and/or disseminate the results of a research project.

Engaging is a term used in the USA by PCORI and the Canadian Institutes of Health Research. PCORI define engagement as meaningful involvement of patients, carers, citizens, clinicians and other healthcare stakeholders in the topic selection, design, conduct and dissemination of research findings. There are six PCORI patient engagement principles: reciprocal relationships; co-learning, partnerships, transparency, honesty and trust. The UK National Co-ordinating Centre for Public Engagement in Research (https://www.publicengagement.ac.uk/do-it) defines public engagement as: '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'.

Devolving is to place decision making in the hands of patients or communities, for example, a community development approach[26].

Consulting is gaining feedback from patients and communities through e.g. meetings, on-line fora, workshops. The role is considered to be relatively passive when compared to 'engagement'

Action Research brings about improvement or practical change. A group of people who know about a problem work together in a *'partnership'* to develop an idea about how it might be resolved. They then go and test this idea. The people who take part in the testing provide feedback on their experiences. It has key tenets[20]:

- Flexible planning – the detailed content and the direction of the research are not determined at the outset
- Iterative cycles with all involved to i) decide what the problem is, ii) decide an action iii) take action iv) learn the lessons from the action v) reconsider the problem and repeat the cycle
- Subjective meanings of those involved determine the content, direction and measures of success of the research
- The research simultaneously improves the situation
- The unique and ever changing social context is taken into account

Co-production means people who use services, members of the public and professionals working together in a '*partnership*' to produce research or service improvement. It is an umbrella term for a concept that means coming together to find a shared solution. 'Co-' can be put before specific research tasks like 'co-design', 'co-build' and 'co-construct'. Co-production[27] covers the whole research process from idea to dissemination of findings in order to change practice.

Co-learning is a term used by PCORI, where the goal is to help patients or other partners to understand research processes. The goal is not to turn patient partners into researchers. PCORI use the term in the context of 'reciprocal relationships', where all research partners including patients learn collaboratively. https://www.pcori.org/sites/default/files/Engagement-Rubric.pdf

Box 3. Overview of how to involve patients in research

A clinician wants to involve patients in a trial of treatment for migraine. Here are steps for involving patients when preparing a research funding application.

1. Understand what patient and public involvement (PPI) is and the different approaches
    i. refer to research funder guidance about public and patient involvement because it varies internationally and is rapidly evolving
    ii. understand how patient involvement differs from patients participating in research
    iii. use language precisely because it varies internationally
2. Find out what research questions are priorities for patients
    i. search the internet for existing work on patient priorities and ask patient organisations
    ii. if patient priorities are unknown, discuss this with your proposed funder and consider how you might fill the gap to progress your research
    iii. prioritise patient-centered outcome measures and find acceptable research methods
3. Identify patients (not your own), charities and/or patient groups to potentially involve as early as possible
    i. consider identifying a professional or lay link worker, perhaps through a charity or a university or hospital patient advisory group
4. Select patients and/or patient groups to be involved in your study
    i. consider equity of opportunity, unheard perspectives and health inequalities
    ii. consider the potential for bias and conflicts of interest
5. Negotiate and agree an approach, tasks and responsibilities at an early stage
    i. consider which approach will add value and rigour to your research
6. Negotiate appropriate funding to pay patients, reimburse expenses, fund activities and staff time to facilitate patient involvement
7. Consider whether training will be required for the proposed roles and responsibilities
8. Consider whether patients or patient groups will 'do' any research
    i. do they have appropriate skills?
    ii. how will they add value and are there risks?
    iii. will they be employed?
    iv. who will mentor and provide supervision?
9. Consider the ethical and research governance implications for involving patients in your study
10. Involve patients in writing the grant application
11. Involve patients to plan future reporting and dissemination of your research

Table 2. Patient participation and patient involvement in research: Methods compared.

Research methods	Qualitative and survey research:patients are study particpants who provide data (text or numerical) on their perspectives to inform research team decisions.		Public Patient Involvement:patients actively contribute through discussion to decisions about research priorities, design, relevance, conduct and governance from study conception to dissemination	
	Strengths	**Limitations**	**Strengths**	**Limitations**
Who contributes? How are they selected to represent the views of larger populations and to meet the aims and objectives of the research?	Sampling strategies are carefully constructed to recruit patients e.g. for specific characteristics, maximum diversity or a representative sample. Usually participants have little experience of research.	Can be resource intensive. Under-privileged groups can be hard to recruit. When to stop sampling is seldom straight forward.	Individuals provide their own patient experience and perspective. Larger charities have well-developed infrastructure, provide equity of opportunity and empower patients to provide meaningful contributions to multiple projects. Active social media sites access a wide range of perspectives. Patients may undertake training for a PPI role.	Selection processes, characteristics and conflicts of interest are seldom reported. Some PPI groups lobby politicians or have influential networks with key decision-makers. Patients can become professionalised, exclusive and immersed in an academic establishment.
How many contributors are required (sample size)?	Variable and flexible in order to answer emergent research questions. Quality standards apply. Small samples may be appropriate if there are qualitative evidence syntheses and few residual uncertainties. Larger representative samples are required where little research exists, to build theory, or if there are multiple uncertainties.	Recruitment problems, lack of resources, limited sites, or convenience sampling can introduce bias. Premature conceptual closure can occur and is more likely when methods tend towards consensus: e.g. focus groups, workshops, Delphi or nominal group techniques.	There is little guidance, so researchers can decide and operate within their available resources. INVOLVE (http://www.invo.org.uk/) state at least two patient contributors attend team meetings.	Individuals cannot fulfil the role of representing all patients and public perspectives. The resources may limit the number and timing of patient contributions.
How do they contribute to each stage: e.g. research questions, design, recruitment, outcome selection, data collection, process evaluation, dissemination, reporting and implementation?	Focused open research questions e.g. about experience of an intervention/ recruitment. Usually at one time point. Qualitative researchers have highly skilled attention to non-verbal communication, contradictions, language and context. Researchers use the findings to inform decision-making. The patient perspective may or may not inform the research design and conduct. Reporting guidelines apply: http://www.equator-network.org	Researcher skills vary. Closed questions asking "what" rather than "how" or "why" will limit data quality, prime responses and can bias findings. Analysis by inexperienced researchers can generate "common sense" or "so what" critiques rather than skilled interpretation.	Range from limited involvement at steering groups to full engagement as a grant co-applicant satisfying the responsibilities set out by the funder. Methods range from informal discussions to partnership research approaches e.g. action research, co-production. Patient partnerships imply recognition, mutual respect, commitment and equity in decision-making. Patients with appropriate skills may be involved in data collection, analysis or interpretation. There are guidelines for using PPI in trials[28] and PPI reporting[29].	Protocols and reports often lack transparency for how PPI is operationalised and impacts on decision making. Grant-holder status may be inappropriate for patients with limited resources and can have opportunity costs, e.g. patients working for a charity may have less time for direct patient support. Views can be undervalued. Alternatively views can lead research in a direction that proves counter-productive. Patients may experience regret or blame.
Ethics, funding and research governance.	Data collection and access require ethics committee approval to satisfy the Data Protection Act and Declaration of Helsinki (http://www.hra.nhs.uk). Participants are not usually paid but can be offered reimbursement for expenses, or a gift to recompense for time or travel. Sponsoring institutions apply research conduct and employment governance to ensure good practice.	Ethics committee approval for study protocols and subsequent amendments are time consuming, and should not be an a priori straight jacket that restricts change. Build flexibility into study protocols for iterative modification of recruitment strategies e.g. if plan A does not work, state plan B and plan C in order to recruit the sample required.	Patient involvement where no data is collected, stored or accessed does not require ethics approval. UK patients are reimbursed for their time (http://www.invo.org.uk/). There are agreements (of varying formality) about what will be undertaken. Confidentiality is expected. Patients are usually independent of academic institutions and funding bodies. Charities usually have their own ethics and governance policies. Build in flexibility for protocol change when using patient partnership approaches like co-production.	If patients recruit, provide, collect or analyse data, ethics approval is required. If patients do research usually undertaken by researchers, the sponsor's research and employment governance procedures apply. Short term study funding can result in productive patient partnerships ending. Patient groups often use social media to seek views, which can pose risks for confidentiality and data protection. Patients should consent to how their contribution to research is reported and declare any conflicts of interest.

improve their understanding of a topic. Focus group discussions or qualitative interviews are audio-recorded and transcribed. Researchers collate, analyse and interpret text data from carefully sampled patients to produce valid new knowledge and generate hypotheses. Qualitative and survey research have systematic methodological quality standards. However, the researcher holds the power and patients may express strong views which may not be reported. In any research, the PPI and the data collection to gain wider patient perspectives can be separate, combined or overlap in some study phases, or they can be completely integrated throughout (Figure 1). Any combination is possible (Supplementary File 2, Example 1). They are often combined and integrated in equitable *partnership* research methods like action research, and 'co-' prefixes to research terms, e.g. co-learning and co-production (Box 2).

Action research, co-design, co-learning and co-production

Action research historically precedes co-production and gathered momentum in the 1940's as a community-led action in research initiative[19,20,30]. The UK National Institute of Health Research (NIHR) who fund research advocate co-production[27] as a method of involving patients meaningfully from start to finish of the research process. Differences in definitions (Box 2) are subtle, vary internationally and researchers may apply the approaches flexibly in practice. 'Partnership approaches' is used in this article as an umbrella term because it acknowledges the changing roles of patients beyond being 'participants' or 'subjects'. Partnership research methods involve patients, clinicians, academics and other relevant stakeholders as equal mutually respected partners in the research team. Being a patient partner implies equal opportunity and equal voice. Equal power in decision-making is sometimes implied, however there are structural and economic power differentials between different types of partner in terms of pay, employment contracts, status and workplace environments. As language is evolving internationally it is more helpful to describe actual patient roles, tasks and responsibilities explicitly rather than use a label for an approach that is open to misinterpretation. For example, co-production[27] may mean consulting patients regularly or patients may actively collect and interpret research data. Terms like 'Participatory Action Research' confuse because the definition of 'participation' in a study means to contribute data, rather than active involvement in research decisions. Partnership research teams decide who has access to participant level data, how to share data securely and how decisions will be made collectively. Partnership approaches can be resource intensive require leadership skills to balance equity of decision-making with a strong scientific rationale. Negotiation skills are required to accommodate different perspectives in order to reach consensus in a timely manner. An important limitation to consider is how the partnership approach is interacting with the intervention: for example action research can become an active intervention component (Supplementary File 2, Example 2).

When starting to design a study about migraine, understand how PPI will add value to the research and which uncertainties about patient perspectives might benefit from additional analysis of patient data from a survey or qualitative interviews.

Find out what research questions are priorities for patients

Many funders require researchers to justify that their research question addresses what is important to patients[31–33]. If a research question is of low priority to the people affected by the condition, or important outcomes are not considered, and/or the intervention in question is considered unacceptable to patients, then further research is wasteful[11].

A starting point for researchers is to find out if patients' priorities already exist for their topic. Many national and international organisations involve patients to identify and publish research priorities specific to a healthcare condition. In the UK, the James Lind Alliance (JLA) specifically identifies and prioritises

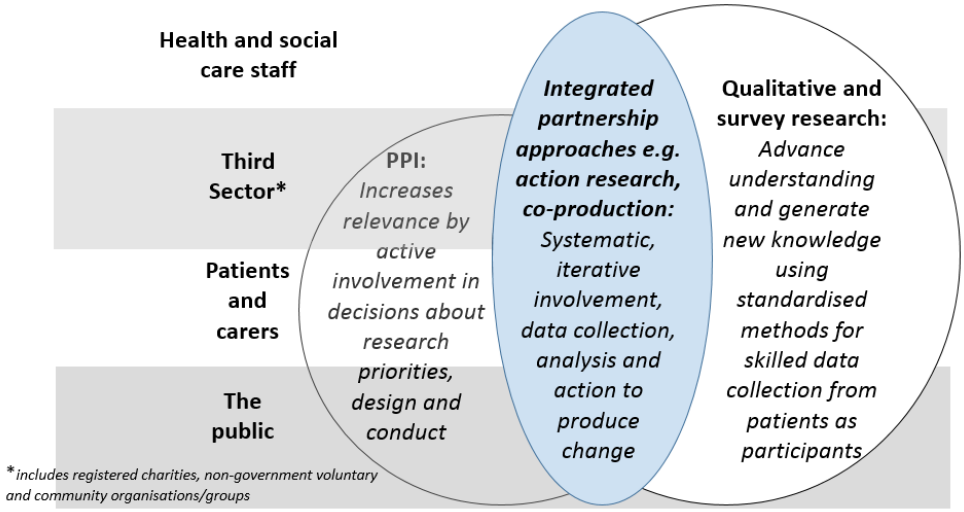

Figure 1. The interface between Public Patient Involvement (PPI), qualitative and survey research across all stakeholders in research.

research questions for funders and there is a register[34,35]. JLA establish Priority Setting Partnerships which involve collaborations between patients, carers and clinicians. The NIHR funds JLA advisors and the infrastructure, but a Priority Setting Partnership is responsible for its own funding. The JLA has a guidebook which provides step by step processes to identify research uncertainties and prioritise a top 10 list for different conditions[36]. Researchers are advised to evaluate how priorities were established and the rigour of processes, as priorities can change with time and some groups may not have had an opportunity to be involved.

If patients' priorities are unknown, and a Priority Setting Partnership is not available, contacting the potential funder to discuss options may be helpful. When researchers plan bespoke methods to prioritise research, it is important to find patients as soon as possible to identify the topic and refine the research question to ensure relevance. For example: work with a charity or a research organisation to conduct an on-line survey (Supplementary File 2, Example 3); advertise and run open public workshops with patients to rank research priorities; or ask participants in qualitative interviews what would make a difference, then construct research scenarios for them to 'think aloud' which one they would prioritise. Once patients have prioritised the research topic and questions, the next step is to prioritise the outcomes that matter, patient-centered outcome measures and identify acceptable research methods.

A first step for a researcher is to search the internet for key organisations and guidelines to see if patient research priorities for migraine are available. If not, a researcher can contact migraine charities and talk to a potential funder to seek their advice.

Identify patients and/or patient groups to involve as early as possible

Researchers are advised to find people to involve and to plan potential roles, responsibilities and tasks for their study as early as possible. Research teams may approach patients through formal patient groups, charities, community groups, University or Health and Social Care patient advisory panels, national directories such as 'People in Research'[37], invoDIRECT[38], patients who are involved in producing guidelines like The National Institute of Health and Care Excellence[39], or through personal recommendation or advertisement. See Supplementary File 2, Example 4. It is usually not considered appropriate to involve patients that members of the research team are currently providing clinical care to[40]. In the UK, InvoDIRECT[38] provides an A-Z on-line resource of organisations, networks and groups that support PPI in health and social care research (Box 1).

Lay or professional coordinators or link people may help and different sources of patients may be used for different purposes. For example, a head office of a patient charity may be invited to nominate a person to join a study steering committee, whereas a local patient group may help to make recruitment materials appealing and easily understood. Participants in a preparatory survey, focus group or qualitative interview may be invited to volunteer for patient involvement in future research. The qualitative research and PPI then become synergistic.

A researcher wanting to study migraine could contact a charity, their University or Health Service patient advisory panel or consult directories of patients who are interested in being involved in research. Invite a patient link worker to join the team who will co-ordinate wider patient involvement.

Decide who and how many patients to involve

As with any appointment, selection criteria for patients based on the research plan are useful to inform decisions. Deciding the number of patients to involve in a study requires careful consideration. Two is the minimum number recommended by INVOLVE[3], however international guidance is less specific. The patient characteristics, skills and numbers will vary according to:

- the study design, e.g. several patients with diverse personal experiences of a health condition may be consulted about which outcomes will be measured in a trial[41]. Co-authors Arthritis Research UK expect patients to be involved in all applications including lab-based early phase research to develop new treatments
- the prevalence of the condition, e.g. it may be challenging to identify two or more patients with rare conditions
- the relevance and reach of a new intervention, e.g. adverts on Facebook for selected postcodes can identify rural and under-privileged urban perspectives
- how much personal tailoring and choice is possible in the design of the research, e.g. two closely involved patients may advise the research team at meetings for a Cochrane Systematic Review, whereas many diverse patient groups may be consulted when prioritising research questions to improve migraine outcomes.

Consider equity of opportunity, unheard perspectives and health inequalities

Equity of opportunity for patients to be involved in research underpins UK guidance. The NIHR standards for PPI[7] provide practical examples for how researchers can offer inclusive opportunities and sustain respectful, productive relationships. There is a danger that patient contributors are atypical, as the more confident and financially secure are more likely to volunteer. It can be easier to involve older, white and educated people, which can marginalise other perspectives. Health inequalities and equity are important when making research decisions[42]. Aim to find patients who represent the demographic of those affected by the condition. It can be challenging to access 'typical' members of the target population for the specific research question[42–44]. See Supplementary File 2, Example 5. An adult or child may be selected to represent their own views[45] or, when the research involves children, vulnerable patients or patients with cognitive impairment, then a guardian, relative or carer may represent the patient's views. A lack of resources can hinder recruiting some patients, such as those from ethnic minorities, the less privileged and less literate. Yet this is important because they tend

to experience lower health status and poorer access to services. For these patients it can feel intimidating to meet researchers and attend meetings in a University. Alternative strategies include researchers going out into the community in order to build rapport and trust with patients on their own turf, which can then lead to discussions about research (Supplementary File 2, Example 1)[46,47]. An outreach model for patient involvement via a link coordinator (professional or lay) can help to access less heard perspectives (Supplementary File 2, Example 5)[48]. A useful guide for getting started and arranging a meeting with patients is available on the INVOLVE website[49].

A charity partner might help a researcher to plan how patients on low-income or from ethnic minorities can contribute to a research study on migraine. Adverts, social media and attractive visual information in local newspapers and chemist shops may help.

Consider the potential for bias and conflicts of interest

PPI in research and political lobbying can co-occur and introduce conflicts of interest with the potential to influence research decisions in ways that have been under-researched[50]. Researchers are advised to consider sources of funding and affiliations of patient contributors, and to re-assess any arising conflicts of interest during their study.

Patients can work with research teams over many years, attend training courses and become a 'PPI methodologist' or expert individual or group. This has advantages and risks. Experienced patients can have an overview of a particular health condition that is invaluable. However, becoming embedded in a research team or an organisation can risk losing the 'eye of the public'[51]. Researchers are advised to consider whether bias due to 'group think'[52] is possible. This is a risk in any established team, for either researchers or patients to become so familiar with the group or clinical area that they lose sight of fresh perspectives. Selecting new untrained patients for a study can highlight researchers' preconceptions and assumptions. However, this also has limitations, as it can be difficult for patients to understand, question and challenge researchers when the language and culture are unfamiliar. Patients who have benefited from or experienced adverse events from a particular treatment can introduce bias. Select patients to balance views, for example patients who have positive and negative outcomes from a new procedure or treatment. It may add rigour to include qualitative or survey research to gain diverse and/or representative patient perspectives.

Throughout all stages of a study, researchers and patients make decisions that need to balance and prioritise evidence, personal experiences and competing values held at the individual, family, organisational, political, cultural and environmental levels. Rigour and quality standards for PPI in research are important to counter critics, as there is still some resistance to implementing PPI[53].

A researcher is advised to consider conflicts of interest and sources of bias, for example links to industry or private companies. Seek to balance positive and negative patient experiences relevant to the study.

Negotiate and agree an approach, tasks and responsibilities at an early stage

Once patients are involved, it is advisable to agree clear boundaries about the scope of the role, specific tasks and responsibilities. Some flexibility is desirable to accommodate unexpected issues that can arise in research and there are grey areas. See Supplementary File 2, Example 6. The approach can be bespoke for each study or for each phase within a study[12,17,44,48,54] and can vary in the level of patient engagement, responsibility and control. Patients can contribute to three key functions: research decision-making; enhancing understanding of patient experience; and advising how to capture knowledge from other patients. For each function, a question to ask is: which method for involving people will add value and rigour? Example 7 (Supplementary File 2) draws on the work of Gamble and Colleagues who have produced a useful list of tips for patient roles in clinical trials derived from a cohort study of 111 funded trials[28].

Be realistic about what will be possible to achieve and the resources required[3]. A template for Terms of Reference is available on the INVOLVE website[49]. Terms of Reference acknowledge the importance of mutual respect, practical communication issues and can be reviewed as the research progresses. Researchers may invite patients to propose ground rules for the length of time required to read and respond to emails and comment on documents, for mutual agreement. It is important for researchers to remember that patients may be managing ongoing health conditions which can be unpredictable. Patients value individual constructive and honest feedback about their contributions in order to learn, gain confidence and maintain motivation[7].

At an early stage a researcher is advised to discuss roles and tasks involved in the migraine study. For example: help to design an appealing patient leaflet, recruit patients, attend project management meetings, interpret findings and present them to lay audiences.

Agree appropriate funding for patient involvement

International arrangements for supporting patient involvement in research vary according to the funding opportunity. It is important for researchers to check current guidance for the funding call they are applying to and budgeting guidance is usually available (Table 1). Negotiate with patients the costs: payment for patient time, any special needs (e.g. childcare, hearing impairment, translation services), training, reimbursement of travel and subsistence expenses. In addition, include costs for staff time to co-ordinate, support, train and facilitate patient involvement. Researchers are advised to spell out to patients the best case and worst case scenarios (e.g. delays to study start and finish), and what contributing to the study would and could involve. Some patients prefer to volunteer, others prefer cash payment or vouchers. Consider patients who are less financially secure. Patients may rely on benefits, part time work or retirement pensions, therefore consider how difficult it is to pay upfront for travel, to

scan travel tickets in order to claim research expenses or to have access to computers or printers to access documents for a meeting.

Preparatory PPI activity prior to submitting the grant application can pose a problem for researchers because funding for this is seldom available prior to a grant. Yet this is precisely when patients can have important impact on the study research question, design and plan. In England, the NIHR Research Design Service will provide small amounts of money to cover PPI at the design stage[55]. Some Universities fund generic patient partnership panels (e.g. 56) to work with researchers who are seeking funding and larger charities can often help[57].

When costing a study about migraine, negotiate sufficient funds to pay for the planned PPI activities, be realistic about the workload and the resources required and consider special needs.

Training for patients involved in research

Providing or offering training may or may not be appropriate depending on the patient role and the purpose of training. Training may be desirable in order to undertake highly skilled roles like reviewing grant applications or sitting on independent trial steering committees. In particular, training in the principles of evidence based medicine, with consideration of where and how patient stories fit in evidence hierarchies may be useful. Example 8 (see Supplementary File 2) provides some training programmes that support patient involvement in research. For patients new to a PPI role, support to develop their abilities and confidence to express their views and question researchers may be relevant. Many universities, research funders and charities provide learning and support activities.

There are many PPI tasks where training is not necessary, where a different perspective is what really matters and patient experience of a healthcare condition is the required expertise. For example, when helping to choose important outcomes or advising on patient information or recruitment strategies, 'untrained' patients may make particularly valuable contributions.

Patients doing research

Traditionally, academics with qualifications, experience and recognised research skills collect and analyse data. However, increasingly patients are helping to recruit participants, collect or analyse data and some UK grant application forms ask about this (Supplementary File 1, Section B). Such questions arguably prime researchers to think that all boxes should be ticked, without considering the implications. Only appropriately trained patients or lay people should undertake research. Shared experiences of a condition can build trust, empathy and a bond which may help to recruit difficult to engage groups, for example children in care[45]. However, attention is required to individual expertise, training requirements, supervision and the scientific rigour necessary to execute high quality research. Patients may do research alongside researchers in partnership research methods[58] and a paradigm of patient-led research is emerging facilitated by social media and digital technologies[59]. INVOLVE has a Patient-Led-Research-Hub to support patients who want to pursue their own research ideas[38].

In the UK, any researcher accessing study participants who are NHS patients or staff requires a letter of access, sometimes referred to as a 'research passport', obtained from the NHS Research and Development offices (Supplementary File 2, Example 9)[60]. If patients or lay people help to recruit participants to research, gain informed consent or collect, share or analyse data from individual or group discussions, qualitative research or surveys, then they are 'doing research' and there are potential governance implications for the sponsor of the research in terms of employment law, ethics, leave entitlement and indemnity. Researchers should not encourage patients to do research because it requires less resource, or because it obviates the need for relatively costly skilled researchers whilst simultaneously bypassing regulatory hurdles. Rather, researchers and patient partners can decide together whether patient researchers are appropriate and beneficial to specific research projects.

Researchers wanting to study migraine may consider the pros and cons of patients doing aspects of the research and the governance issues.

Working together ethically

Consider how to work with patients ethically. PPI can be empowering for individuals and communities, but there are tensions and risks, including exploitation[25], and the burden and resource implications can be considerable[10]. Some ethical principles for researchers to consider when involving patients in research include:

- avoiding discrimination, undue persuasion, excessive burden or creating a sense of obligation to be involved in the study
- the distribution of power in research
- valuing patient contributions and fair financial compensation
- conflicts of interest, research integrity and respect for intellectual property
- the confidentiality of data and protecting anonymity of research participants
- advancing science through honest and accurate reporting.

INVOLVE[3] states that UK ethics committee approval is not required when patients advise research teams, prioritise research questions, make choices relating to design, share decision-making or disseminate research findings. However, there can be grey areas particularly in relation to defining 'data collection'. NHS or University Ethics committee approval is required in the UK if personal information, i.e. data as defined in the Data Protection Act[61], is collected, shared and stored for future analysis and reporting. For iterative partnership research approaches like co-production, the current ethics committee processes create many challenges[62]. Researchers can request informed consent from participants to share anonymised data with patient partners, so that they can be involved in analysis and interpretation as members of the study team.

There are international differences in requirements for research ethical and governance approvals, and particular challenges with digital health research[25] which are beyond the remit of this article. New EU General Data Protection Regulation[63] commenced in May 2018, and requires transparency about the source of personal data, the purpose and who data will be shared with.

Audio-recording of PPI meetings in order to write accurate but not verbatim minutes, does not require ethics committee approval. However, it does require at least verbal consent from all present at the start of the meeting and the recording should be destroyed as soon as the minutes are agreed. People should receive forewarning of the intention to audio-record, know the purpose, what will happen to the recording and to the content, and be able to object or withdraw. If audio-recordings are stored for longer than is necessary, transcribed verbatim or if there is an intention to report or publish potentially identifiable quotations or content arising from PPI activities, then ethics committee approval is required. Ethics committees have lay committee members, who consider the ethical issues relating to patient involvement.

A researcher wanting to study migraine should consider the ethical issues when involving patients in the design and conduct of their study. Consider patient burden, equity and power, fair and respectful arrangements, confidentiality and the purpose, processes and consequences of any data collected or stored.

Involve patients in writing a grant application

Patients sit on research prioritisation committees and funding panels, alongside clinicians and academics, to decide which research is commissioned and which grants are awarded. See Supplementary File 2, Example 10. Many UK funding panels expect to read convincing and meaningful accounts of how patients have had an impact at key stages: preparatory work to inform the planned research; writing the application form particularly the lay summary; and the proposed PPI activity during the study. Expect to be challenged if PPI appears tokenistic. It is important to consider the trade-offs between specifying a plan for PPI in a research protocol and building in some flexibility for change as the research progresses. This may be challenging in countries where regulatory approvals for amending protocols is time consuming.

Patients can help researchers to write the whole grant application in an engaging, easy to understand language. The lay summary is often one of the first sections in a grant application that funding committee members read to gain an overview of the study. Reviewers like to understand exactly what study participants will experience from start to finish. Describe PPI clearly so that the reader understands who, why, how many, how often, what methods and what impact patients have already had on the grant application and will have in contributing to future research decisions. For example, decisions about recruitment methods, intervention delivery or components, which outcomes will be primary or secondary and how to collect data. It helps to use language precisely and to understand how *involving, participating, collaborating, consulting and engaging* with patients in research differ (Box 2).

A patient helping to write and edit a grant application can make it clear what will happen to patients who participate and how patients will be involved from study conception to dissemination of findings.

Plan future dissemination of findings

Patients can advise on how research might have an impact on health and health care beyond an academic audience. They often have in-depth knowledge of their condition and of on-line sources of information beyond that of academics and clinicians. They can help to write reports, blogs or summaries of findings creatively. See Supplementary File 2, Example 11. Offering participants a lay summary of the research findings is good practice. 'Patients Included Charters' provide accreditation for involving patients in conferences and in journal publications[64] and GRIPP2 PPI reporting guidelines[29,65] are available. Involvement of patients and the public is a critical component in successful implementation of research findings into healthcare, although evidence for best practice is limited[3,66,67].

The grant application for a study about migraine may propose a public event with a charity to present the results of the study. Researchers and patient partners may give joint talks. Small group discussions with migraine patients can suggest ways to spread the news and change care.

Conclusion

This article provides a starting point for researchers and patient partners who are planning to seek funding for research. There is no current international consensus on best practice or terminology and guidance is evolving across countries and research disciplines. A crucial distinction when gaining patient perspectives is between patient *involvement* in research and patients *participating* by providing data in surveys, qualitative interviews or group discussions. The ethical governance implications differ particularly regarding data protection.

Researchers and patient partners can choose a wide range of different approaches to PPI and each study will require consideration of the optimal approach. Rigour is needed because patients' lived experience and persuasive narratives can influence important research decisions and the outcomes are not always predictable. Evidence is needed about how different methods of involving people can improve research decisions, healthcare outcomes and impact. A more collaborative and reciprocal partnership approach with patients has the potential to ensure that research undertaken matters to a wider tranche of society and involves those who stand most to benefit from it.

Key messages

Important questions for researchers about including PPI in their research:

- How can I find people in society (patients, patient groups, carers, the taxpaying public, lay organisations) who can make important contributions to research design, conduct and dissemination?

- How will PPI help me to access the perspectives of those who the research potentially will impact on?
- How can different approaches to involving patients as consultants, collaborators or partners improve the relevance, quality, future implementation and sustainability of research?
- How can patients contribute to three key functions: research decision-making; enhance researchers' understanding of different perspectives; and knowledge capture?
- How can PPI, qualitative research and surveys of patient opinion be optimally combined?

Data availability

No data are associated with this article.

Author information

PH wrote the first draft. All authors have contributed to and approved the final version. PH and IB are members of the NIHR/HTA Commissioning Board and General Board respectively and AOC was a member of the NIHR Programme Grants for Applied Research panel 2007–2017. JLD is an emerita NIHR Senior Investigator and was previously on the NIHR HTA Commissioning Board, NIHR HSR Board and CRUK Population Health Board. JT has lived with rheumatoid arthritis for over 30 years and is also a carer for a brother with schizophrenia. She has been involved in PPI for 7 years covering the whole spectrum from basic science to applied health services research. Her background is in higher education and she works part time for the Open University. CM is the research manager for Arthritis Research UK, a registered charity in England and Wales no. 207711, Scotland no. SC041156. PH and AP have worked as a General Practitioner and as a Physiotherapist respectively. AP is an associate editor with Cochrane Stroke and has received funding from Cochrane Training to synthesise evidence relating to PPI in systematic reviews[68]. PH has been a chair and deputy chair of a research ethics committee. PH is guarantor and affirms that the manuscript is an honest, accurate, and transparent account of the analysis reported.

Competing interests

No competing interests were declared.

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Supplementary material

Supplementary File 1: Section A, Additional terminology and international information relevant for patient and public involvement in research at the funding application stage; Section B, Questions asked by UK National Institute for Health Research (NIHR) on funding application forms.

Click here to access the data.

Supplementary File 2: Examples of different approaches to incorporating patient and public perspectives into research design and conduct.

Click here to access the data.

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Open Peer Review

Current Referee Status: ✔ ? ✔ ? ✔

Version 1

Referee Report 12 November 2018

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✔ **Clare Jinks**

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Thank you for the opportunity to review this paper. The aim of this paper is to discuss how researchers can involve patients when applying for research funding, and to outline opportunities and pitfalls. It uses a combination of knowledge about this process from literature and the authors' wide ranging experience.

This article provides a general background to patient and public involvement in research. The article very usefully includes in Box 1 and Table 1 current resources and guidelines that are available. The information is very comprehensive and relevant to the aims of the article - which is how to to incorporate PPI into the design and conduct of research. This overview is unique and in my view will be widely used. However can it be moved to later in the article? The tables do break up the flow of the paper and break up what I think is an important section about the difference between PPI and patient participation in research.

The section on the difference between patient involvement and patient participation in research is much needed as PPI is often conflated with qualitative research. This is still a common problem. I wondered if Figure 1 could come earlier, or if Box 2 can show the differences more explicitly instead of providing a list of definitions, can the differences be displayed side by side, using maybe column headings of differences and similarities. This will enable the reader to access the important distinctions more easily.

On page 10 in the section about action research, co-design and co-learning there is a sentence about how a partnership approach may interact with an intervention. I think this is the first time intervention studies are mentioned as previous text has stated that patient participation in research may be through interviews, focus groups and surveys. I wondered if this could be expanded to include that these can be undertaken in the context of trials, as then the text would align better with the material in the supplementary documents.

There are a lot of examples in the supplementary files and this can be quite difficult to navigate.

I have an observation on how the steps are characterised. At first I thought the text in italics are the recommended steps for researchers? For example on page 11 the first step for researchers outlined is to search the internet for organisations and guidelines to see if research priorities have been established in their area of interest. There is however some advice given earlier (written in italics) that recommends understanding what PPI will add and which uncertainties about patient perspectives may benefit from more exploration.

Then I realised that the steps are characterised in the blue subheadings. These are a mix of 1. advice (e.g. understand what PPI is (this is a conceptual issue)), 2. questions (e.g. how does PPI differ from patient participation?) and then 3. labels for different issues (e.g. patients doing research, which is a practical issue). Could there be more consistency across the paper? I wondered if it would work if they were all translated into specific statements / recommendations particularly as the aim is to provide guidance on how to incorporate PPI.

Related to the above - I wondered whether the section headings of "how does PPI differ from research participation" and action research, co-design, co-learning and co-production" should be at the same level as the others. They are very important - but I think are fundamentally about understanding what PPI is.

The text in italics is also a mix I think of statements of principles - e.g. seek to balance positive and negative views, rather than a statement of how to do that (the title of the paper is how to incorporate PPI....)

The section on how many patients to involve is again really helpful as this is the question that people often ask. This is usually asked in the context of "representativeness" of the patients' views. Can this section be expanded - with some examples of what teams can say when the "representativeness" of their patient advisors is questioned. It links to a later section on bias and conflict of interest.

The authors recommend that researchers select patients with balanced views. It is difficult to know what view people hold at the outset. Can this recommendation again be expanded upon - with ideas about how you may do this, or how you may get fresh perspectives over time.

Towards the end there is a heading about involving patients in writing a grant application. This made me realise that the advice spans both the process of PPI at pre-award and then outlining what to do post award. I wondered therefore whether this section should come earlier. PPI advisors will have an input into the design of the PPI throughout and the research design throughout, and therefore impact on decision making about earlier topics in the article.

I think this is a really valuable paper in providing such a comprehensive overview of resources and guidelines for PPI and in highlighting the key challenges of good PPI practice. My suggestions are really about structuring the content to make it more accessible to the reader by highlighting maybe two aspects. First the need to understand what PPI is and is not. Second the practical issues - how to do it.

Is the topic of the opinion article discussed accurately in the context of the current literature?
Yes

Are all factual statements correct and adequately supported by citations?
Yes

Are arguments sufficiently supported by evidence from the published literature?
Yes

Are the conclusions drawn balanced and justified on the basis of the presented arguments?
Yes

Competing Interests: No competing interests were disclosed.

Referee Expertise: I am an applied health services researcher (musculoskeletal conditions and long term term conditions) with an interest in PPI practice and research

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Referee Report 02 November 2018

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? **Kathryn M Sibley** [1], **Patricia Roche** [2]

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Thank you for the opportunity to review this manuscript. This article gives an overview of steps research teams and patient partners can take when working together to prepare a research funding application. We believe a guide for inclusive and meaningful involvement of patients and public partners, particularly in the planning stages of research, is a highly useful and timely resource for health researchers. We thought it was appropriate that the authors included a variety of resources from different continents, and appreciated the attempt to summarize the varied (and often confusing) terms and concepts that exist around patient and public involvement. It was great to see involvement of patient partners in authorship of the paper. We also wonder if patient partners have been involved in providing review comments as well.

We did struggle with the overall flow of the article, due to its non traditional structure, thereby making it challenging to follow for the reader. Despite the valiant attempt to describe differences in terminology around patient and public involvement in research and key concepts that need to be considered for meaningful patient and public involvement, terms are at times dispersed, seem to be used interchangeably throughout the article, and do not follow a step-by-step order that would be expected from an overview of ‘steps’ in a process. In addition, many key considerations that should be made prior to engaging patients or members of the public are left to the end of the article (such as training for patients involved in research, or working together ethically). In general, the article operationalizes PPI into a series of steps and although mentioned at some points later in the article, the theory and principles of equity and social justice that are critical to meaningful and inclusive engagement are presented later in the article. However, to ensure researchers are incorporating these principles, we suggest they be introduced sooner and have a more central role as the lens through which PPI is viewed. We feel it would be more beneficial to begin with terminology and key concepts, and then provide an overview of the subsequent steps involved in engagement.

Given that the article appears to be an attempt to synthesize information and provide an overview of the steps to take when involving patients and the public in preparing a research grant application, a clear description of the methods used to gather, sort, and summarize the sources and information contained in the article is missing. Such information is critical for demonstrating the strength and rigor of the recommendations. Although we recognize this is an opinion article that is not intended as a formal synthesis, we feel that including a description of the literature review methods would provide a landmark for others who may seek to undertake a full synthesis in the future.

The tables and boxes included are excellent resources for researchers seeking to learn more about PPI and how it is done in different contexts and regions. However, aside from the definitions, their placement

may be better suited to the supplementary material, rather than embedded within the article itself (especially Table 1, as it is in landscape orientation making it difficult to read in digital form).

As we have heard from our stakeholders and patient and public partners, language is a critical aspect of ensuring that PPI is successful, inclusive, and respectful. Many of the decisions that should be made through dialogue with patient partners are presented as decisions for the research team to make and share with patient partners. We recommend directing researchers to dialogue with patient and public partners about topics such as ground rules, preferences for compensation, preferences for feedback, and other matters in which patient partners are involved, in order to ensure that decision-making power in engagement activities is equally shared. A specific example is on page 12 under the heading “Agree appropriate funding for patient involvement”, where the authors state that researchers “Negotiate with patients the costs: payment for patient time, any special needs…” and “Researchers are advised to spell out to patients the best case and worst case scenarios…” – we caution to consider carefully how language can challenge or perpetuate power differentials that often tend to place the researcher above the patient as having more expertise and education. We suggest that terms like ‘discuss’ and ‘dialogue’, in place of ‘negotiate’ and ‘spell out’, can help work towards equality and mutual respect in the research partnership. Some specific points the authors may want to address are detailed below.

Page 3:
Given that the concept of PPI is central to the article, a definition (or a discussion of the variability between definitions) should be one of the first points of discussion.
The authors state that “PPI includes patients, potential patients, families, carers, patient groups and members of the public who use or have access to health and social care services”. We would suggest PPI also includes those who may be currently unable to use or access health and social care services.

The authors discuss ‘patients’ including “people who do not describe themselves in this way”, an important consideration for ensuring inclusive and meaningful engagement – however, we feel the discussion of why people may not describe themselves as ‘patients’ (for example, medicalization of people with disabilities, stigma attached to living with a mental health issue).
Box 1:
It would be of great interest to us to know how the resources listed in this box were chosen and why. We also believe that given the iterative nature of many online resources and the frequency with which URLs change, it may be best to avoid including hyperlinks for specific documents, many of which will likely become defunct within a year’s time.
Box 2:
Under ‘some acronyms for involving people in research’, it is unclear why this particular handful of terms was chosen, yet we do not see common acronyms such as PE (patient engagement; https://bcsupportunit.ca/patient-engagement-methods-cluster) and PAR (participatory action research[1]) .
The section of definitions appears to lack structure or hierarchy and is confusing in its presentation. Rather than clarify terms, this box seems to further complicate the differences in language used. Specifically, ‘partnership’ in engagement should also include participatory action research and community-based participatory research[2]; devolving would also seem to fall under ‘partnership’; ‘consulting’ is described as “relatively passive when compared to ‘engagement’”, but still falls within the realm of ‘engagement’.

Box 3:
This overview is an excellent concept and will be highly useful to research teams looking to engage with patients in their work. However, we feel that there may be some information not found here that may be useful. This includes a step for assessing capacity of both research teams and potential patient and public

partners to meaningfully engage – for example, determining the willingness of the research team to change directions based on patient and public partner input; determining what resources are available to support engagement activities; and determining the underlying purpose for engaging.

Point 3 indicates researchers should identify patients to involve "as early as possible" – this may be unclear to those unfamiliar with the PPI process, and should be more explicit – i.e. engaging before the research questions and methods have been determined – something we do not see mentioned elsewhere in this article.

Under 3.i, it is suggested that researchers consider identifying a "professional or lay link worker", however many research teams will not (for whatever reason) choose to hire an outside professional to assist with engagement activities, and will embark on it themselves. For this reason, we feel it is critical to mention the need for reflexive practice and trauma-informed approaches for those who are not familiar with these approaches and their need when engaging with people with lived experience of health issues, and the potential retraumatization associated with sharing those experiences.

Under 5, there is a minor grammar error – "agree an approach" should read "agree *on* an approach".

Under 6, we believe the use of language such as 'negotiate' may perpetuate power imbalance between research teams and patient and public partners. This language posits researchers and patients on opposing sides, when perhaps what the authors intended to suggest with 'negotiate' was a method of working together to determine appropriate funding and compensation – with which we agree. Perhaps using a term such as 'researchers and patient partners should work together' instead of 'negotiate' would be more appropriate. In relation to this point, it seems that a key component missing from this article's discussion of compensation for patient and public partners is determining the preference of patients and *actually asking them*what they want in terms of level of involvement, supports, payment, etc. Similarly, under 8, the authors suggest that researchers "consider whether patients or patient groups will 'do' any research" and subsequently assess their skills, ability to add value, status as employees, and mentorship/supervision – we again suggest that these considerations should be made *with* patient and public partners, rather than decided for them.

Page 10:
From the title of the section "Action research, co-design, co-learning and co-production", it seemed as though the authors would be providing a discussion of these different principles, but instead was a discussion of the differences between definitions that should probably have been included at the beginning of the article. We would suggest moving this information to before the 'steps' of involvement are discussed, and potentially changing the heading since it is somewhat misleading given the content of the section that follows. Additionally, the first sentence discusses the historical context of 'action research', yet this is the only mention of the chronology of terms and seems somewhat out of place. Without discussion of the origin of other terms mentioned in the article, we would suggest that this is unnecessary to the reader's understanding of concepts.

Under "Action research, co-design, co-learning and co-production", there is a minor grammatical error – in the sentence "Equal power in decision-making is sometimes implied, however there are structural and economic power differentials between different types of partner in terms of pay…", the word "partner" should be pluralized to read "partners".

The authors have brought up a critical consideration regarding power differentials between patients, however the discussion of these and other systemic power imbalances seems to be somewhat lacking in

the article, particularly regarding the dynamics of relationship-building in research partnerships, where researchers may hold more 'power' and need to engage in reflexive practice to understand how this impacts the process of engagement. We also recommend a discussion of safe spaces, a key consideration for addressing power imbalances in research partnerships. As a reference, we would suggest reviewing the 2017 BMC Health Services Research article by Shimmin et al.[3].

In the second column on this page, the sentence "Partnership approaches can be resource intensive require leadership skills…" appears to contain a grammatical error and should perhaps read "Partnership approaches can be resource intensive and require leadership skills" or "Partnership approaches can be resource intensive, requiring leadership skills".

The authors mention "negotiation skills are required to accommodate different perspectives in order to reach consensus in a timely manner", however we are not sure that this aligns with the principles of meaningful and inclusive PPI in research – though consensus and timeliness are obviously of importance to researchers, we are not convinced that these are central tenets of engagement, and would argue that respect for patients' stories and lived experiences should be valued over 'timeliness'.

Under "Find out what research questions are priorities for patients", the authors state that "If a research question is of low priority to the people affected by the condition, or important outcomes are not considered, and/or the intervention in question is considered unacceptable to patients, then further research is wasteful". Although we do not disagree, this is a bold statement that we would suggest tempering, given that basic, fundamental research is often of low priority to patients, and may not have a direct intervention for treating disease or improving health, though it often serves as critical foundations for future discoveries.

Figure 1:
The figure demonstrates the people involved in different types of activities. It seem as though the integrated partnership approaches are intended to be positioned as the 'overlap' between PPI and qualitative and survey research. Generally, we found this figure is somewhat difficult to interpret and aesthetically unpleasing. We also question the exclusion of health and social care staff from PPI, and wonder why people in these roles would not be able to participate in PPI.

Page 11:
In determining patient priorities, we are pleased to see mention of JLA, an important methodology. We also think this would be an excellent opportunity to introduce the Patient-Led Research Hub, mentioned later in the article.

The use of survey methodology as an example of identifying patient priorities seems overused – there is a lack of discussion of other potential priority-setting methods (such as patient journey mapping and digital storytelling). This is particularly important for researchers who are new to the concept of PPI who would benefit greatly from information about alternatives to the survey methodology.

Mention of hard-to-reach groups should be mentioned initially under "Identify patients and/or patient groups to involve as early as possible", to ensure it is at the forefront of researchers' minds when considering their recruitment strategy.

Page 12:
Under "Consider the potential for bias and conflicts of interest", we would recommend mentioning that this should be a consideration for discussing ground rules and/or terms of reference. This would also be a

good time to mention that the research team and patient partners should discuss together what to do in cases of potential conflicts of interest, and how those will be addressed in the context of the research partnership.

Within the heading “Negotiate and agree an approach” there appears to be a word missing, such that the heading should read “Negotiate and agree *on* an approach”. Additionally, in the first sentence of the paragraph under this heading, “Once patients are involved, it is advisable to agree clear boundaries…” should read “Once patients are involved, it is advisable to agree *upon* clear boundaries…”.

In discussing the ‘scope of the role’, it is also important to note that the role of the researcher in the partnership should also be discussed, and this should be indicated in the article.

The authors state that “Patients can contribute to three key functions” – we would argue that patients often contribute to other functions such as collecting data, interpreting results, and informing knowledge translation activities, to name a few.

The authors state that “Patients value individual constructive and honest feedback about their contributions in order to learn, gain confidence and maintain motivation.” Although this statement may be true, the indicated reference is a link to a NIHR site declaring the development of national standards for PPI (from March 2017). This statement in particular should be supported by a reference, or replaced with the suggestion that research teams discuss with patient and public partners how they want to receive feedback and what they need to learn, gain confidence, and maintain motivation.

The heading “Agree appropriate funding for patient involvement” seems to be missing a word, and should probably read “Agree *on* appropriate funding for patient involvement.”

The sentence “Negotiate with patients the costs: payment for patient time” should also include expertise, a major contribution of patients to the research process, such that this part of the sentence should read “payment for patient time and expertise.”

Page 13:
The inclusion of consideration for patients not having to pay upfront for travel and having access to computers and printers is critical and we commend the authors for including it.

In discussing the steps for PPI in grant development, discussing how to budget for these early activities would be appropriate and useful for those reading this article. At the very least, a reference to an existing budget tool (such as INVOLVE’s cost calculator - http://www.invo.org.uk/resource-centre/payment-and-recognition-for-public-involvement/involvement-cost) would be appropriate and highly useful.

Under “Working together ethically”, it would be helpful to include references for researchers to explore these concepts in more detail – how to ensure distribution of power in research, perhaps guides to reflexive practice questions (such as Shimmin et al.’s article), guidelines for valuing patient contributions and fair compensation (such as those produce by the SPOR National Disease Networks).

Page 14:
The authors mention “Audio-recording… does not require ethics committee approval” – we are unsure whether this advice is accurate for all potential jurisdictions, and would suggest including a note about inquiring with the readers’ own regulatory bodies would be appropriate here.

The authors warn “Expect to be challenged if PPI appears tokenistic”, but provides no guidance for what this means and how to avoid it. We would suggest including more detail and/or a reference.

The authors state that “It helps to use language precisely and to understand how *involving, participating, collaborating, consulting and engaging* with patients in research differ” - this statement gives the impression that these concepts are mutually exclusive, when in fact they are overlapping and intertwined. Perhaps this could be restated as “how *involving, participating, collaborating, consulting and engaging* with patients in research differ, and what the overlap between these concepts is.”

In the Conclusion, the authors call for evidence about the engagement process, and we would suggest they may also want to touch on the potential to have patient and public partners included as participants (in evaluating the engagement process), and should discuss this when issues around ethics are discussed (page 13) and/or when comparing partners and participants (page 3).

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Is the topic of the opinion article discussed accurately in the context of the current literature?
Yes

Are all factual statements correct and adequately supported by citations?
Partly

Are arguments sufficiently supported by evidence from the published literature?
Partly

Are the conclusions drawn balanced and justified on the basis of the presented arguments?
Partly

Competing Interests: No competing interests were disclosed.

We have read this submission. We believe that we have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however we have significant reservations, as outlined above.

Referee Report 16 October 2018

https://doi.org/10.5256/f1000research.16517.r39334

Sally Crowe [1,2]
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Thank you for the opportunity to review this article – which I think is an important mile stone publication in the business of patient and public involvement in the activity that prepares and contributes to research applications. This is a timely and useful review of the literature and the experiences of the research authors, one day we might look back and realise that investing in this stage of research yields benefits and savings in research down the line.
I was interested in the reasons for the authors writing this article and was not surprised to see that it was about the conflation of qualitative research and patient and public involvement in research. This article helpfully, and in a practical way helps to 'de couple' the most pertinent issues in this domain.
The use of an illustrative example in migraine research helps to bring the review of evidence and discussion in each section to a useful and practical conclusion for the reader, and could be easily re interpreted for their own research context.
It is also a strength in this article that research is considered as both primary studies and research synthesis and reviews, both of which require careful patient and public involvement
Page 3 – in the section *How does patient involvement differ from patient participation in research?*
"the context and outcomes from listening (to patients) differ. PPI means that researchers are in a continuing and reciprocal relationship with patients and make decisions with them about the research. In qualitative research, researchers listen to patients in order to improve their understanding of a topic". This is probably the most helpful sentence I have read in a while! The further discussions about the choices of researchers that may or may not include qualitative perspectives in their analysis further underlines that in PPI the power dynamic is different and it is an important difference.
Figure 1 is helpful
I have a problem with the word iterative – and would suggest a plainer language option especially as the rest of the language used in the diagram is of the non-research variety.
Table 2
I struggled with this table maybe because I am not a researcher – I would prefer to see the differences between the two rather than the strengths and limitations of each but can appreciate that for researchers making choices this might be very helpful information and analysis.
Boxes 1 and 2
For an article such as this I think that the *contents of Box 2* are more helpful for readers untangling what is meant by PPI in pre funded research and *would put Box 1 as a supplementary file* – this information is more easily found for a curious researcher and Box 2 really adds value to the article as a whole as authors have collected the (sadly) rather large collection of terminology used in PPI and sought to differentiate it. I like the fact that the authors have stated and used their preferred terminology and articulated the reasons for their choice (partnership approaches).

Table 1
I imagine this could be immensely useful to researchers and it is a useful comparison tool but positioning in the middle of the narrative is a shame I think as it breaks up the flow of reading.

Find out what research questions are priorities for patients
I think that there is a step before initiating priority setting exercises that encompass PPI – increasingly there are published accounts of priority setting that may or may not include the perspectives of patients and the public. There is also an emerging checklist to support the quality assessment of these accounts and specifically the degree of PPI in them

- Tong A., Sautenet B., Chapman JR., Appraisal checklist used in a systematic review of priority setting partnerships in health research (Currently being reworked as a priority setting reporting checklist and will be renamed REPRISE Checklist).

Consider equity of opportunity, unheard perspectives and health inequalities

This is a particularly important and pertinent section and offers practical advice and ideas for researchers, especially the use of outreach models that offer more scope for addressing these issues more directly, but may put researchers out of their comfort zones.

Consider the potential for bias and conflicts of interest

This feels an underwritten first paragraph. It feels more important to instigate transparency and declaration of financial and other interests *for all* research participants – than the more in-depth description of the 'experienced patient?'. Additionally some text about how to manage these conflicts of interests in patients/public may really help readers address this issue.

Agree appropriate funding for patient involvement

There are important considerations here especially around equity of involvement – its good to see these spelled out.

Training for patients involved in research
I was very pleased to see this in the text for this section "For patients new to a PPI role, support to develop their abilities and confidence to express their views and question researchers may be relevant." A much under-appreciated aspect of PPI, I would suggest that it doesn't just apply to those new to a role who may find it harder (as am embedded part of the research team) to challenge the research orthodoxy…

Also the training needs to be two way – for research teams as well as involved patients and the public.

Key messages Important questions

I like these but I would include a challenging first question to researchers – 'Why do they want to do PPI?' I think that there is an important aspect of self-discovery in PPI in research and it helps to understand self/organisational motivations to doing this preparatory work. These reasons may encompass rational or outcome-based reasons (which the current list addresses) but it is also important to understand motivations on a human and relational level.

Is the topic of the opinion article discussed accurately in the context of the current literature?
Yes

Are all factual statements correct and adequately supported by citations?
Yes

Are arguments sufficiently supported by evidence from the published literature?
Yes

Are the conclusions drawn balanced and justified on the basis of the presented arguments?
Yes

Competing Interests: No competing interests were disclosed.

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Referee Report 02 July 2018

https://doi.org/10.5256/f1000research.16517.r35143

Kristin Liabo
National Institute for Health Research (NIHR) Collaboration for Leadership in Applied Health Research and Care South West Peninsula (PenCLAHRC), Medical School, University of Exeter, Exeter, UK

This article gives a comprehensive overview of the involvement of patients, carers and members of the public in health research. It is well written and addresses some important thorny issues in regards to involvement. The article is likely to be useful for researchers new to involvement. Also, as someone who has been facilitating involvement for some time it is very useful to see all this information pulled together. Partly due to the comprehensiveness, I suggest some of the information in boxes and supplements could be cut down. The sheer volume of information might confuse novice readers.

For example, the authors point to the many different and overlapping terms used in this field. For a new author it might be more helpful if the information about all these terms was reduced so that the article focuses on the substantial differences rather than the terms used. I found boxes 1 and 2 quite overwhelming to read through at the beginning of the article. Perhaps it would help if they were supplementary files instead of in-text?

Another example is the definition of ‘participating’ – I found the introduction to participatory action research here quite confusing because this can be a study design where involvement and participation happen in tandem or are intersected.

Overall, I would have liked to see less of the detailed information on various websites and terms, and more incorporation of the thorny issues, e.g. where research and involvement intersect, and discussion about what we can do about this. It is these aspect of the article that are most interesting, in my view. But the attention to this would depend on the purpose of the article.

Some other comments:
In Box 3, which gives an overview of the involvement process, I would suggest not using the term bias, because this is commonly used for samples. I agree with the point made – and it is important as often ignored in involvement guidelines – but I think it would be better not to use bias and instead say something about whether the topic of the research is contentious amongst different groups of patients (or something to that effect).

Also in Box 3, bullet number 7 mentions roles and responsibilities. I would suggest the agreement of these needs to come much earlier in the process. For example, this could come under point 1 – when the researcher familiarises themselves with involvement. What roles would they like patients, carers or members of the public to have?

I really like Tables 1 and 2 – these are super useful overviews. I am not clear what purpose Supplement 1 has, this relates to my previous point on the brevity and detail of information.

In Supplement 2 I find some of the examples lacking in purpose and clarity beyond saying that involvement can happen. I don't find that most of the examples provide new information that isn't available elsewhere in many different formats. However, some examples are very interesting and could merit more space. These are: Example 2 about the cautionary tale – this is a new point that I have not heard before; Example 6 which gives a very interesting example of when qualitative research and involvement overlap (again, would merit more discussion); Example 9 which is very brief but points to how protectionist policies can exclude people from participating (a common reason for not involving children, young people, people with disabilities, the frail elderly and other vulnerable populations) – I don't think this has been considered in-depth by policies intended to increase participation; Example 10 is good on details on *how* researchers can work collaboratively with patients/carers/members of the public and will be of interest to people looking for new involvement ideas.

You describe patients as primarily fulfilling three functions when they are involved in research: research decision-making, enhancing the patient perspective, how to capture knowledge. I suspect many involved patients/public advisors will object to this as being too limiting. In my own work I have seen at least three additional kinds of input: 1) public advisors helping researchers plan involvement in their research, 2) public advisors helping with dissemination and collaboration strategies (recently a public partner pointed out that the researchers had not asked for a letter of support to a very key national charity which could really help with dissemination), and 3) what I'd call 'hidden or obvious talents'. Examples of the latter are patients drawing on their previous careers or hobbies, or other talents for seeing new aspects of an established research method. A parent carer we work with decided to use VideoScribe to disseminate some research she had initiated, she then presented about this software at a research seminar, and the research programme bought the software as a direct result of hearing her talk about it. In this example the function of the involved parent was to influence the dissemination and communication strategy of a whole programme of work.

I hope these comments are useful.

Is the topic of the opinion article discussed accurately in the context of the current literature?
Yes

Are all factual statements correct and adequately supported by citations?
Yes

Are arguments sufficiently supported by evidence from the published literature?
Yes

Are the conclusions drawn balanced and justified on the basis of the presented arguments?
Yes

Competing Interests: I am employed by a research grant to facilitate the involvement of patients, carers and members of the public in their research. This means I have a vested interest in this activity being improved and promoted.

Referee Expertise: Patient and public involvement in research

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Referee Report 25 June 2018

https://doi.org/10.5256/f1000research.16517.r35144

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A helpful article. A few recommendations that will help improve the accuracy and help avoid any confusion.

One step that is missing I think is 'Preparing the research team for PPI'. Support and training are mentioned in relation to the public but this also applies to the research team.

p1. 'The research methods of PPI...' I don't think 'methods' is the right word (eg 'informal discussions' is not a research method). Perhaps 'techniques' or 'The ways in which patients and public are involved in research...'

p3. I think the sentence in the first para beginning 'In the UK, INVOLVE states...' should come at the end of that para.

p3. 'Steps for how etc'. Are these steps? Or are they 'Issues to consider'?

p3. 'Understand what patient and public involvement is'. A more accurate heading would be 'Understand your rationale for patient and public involvement.' And in the next two sentences you mention 'theory'. I don't think these are theories - I think it is about 'understanding the rationale or motivation for patient and public involvement.'

p3 'How does patient involvement differ from patient participation?' The only step that is posed as a question - I'd change to 'Understanding how patient involvement differs etc'. And it would be worth adding in again to this section (I know it's already in elsewhere) INVOLVE's distinction between involvement and participation.

p7 The list of acronyms includes both terminology and organisations. Confusing. Take out the organisations - if they're in the main body of the article then they should be written in full anyway.

p11 'A first step....'. I'd reword to make less like an instruction and consistent with the rest of the article. So 'A researcher could search the internet etc. Another approach would be to contact migraine charities etc.'

p11 Para beginning 'Lay or professional coordinators or link people...' Consider explaining what these terms mean. And replace 'whereas' with 'and'. Lose the sentence 'The qualitative research and PPI become synergistic' - it confuses the point being made in this section and I'm nort sure it's accurate.

p11 'Decide who and how many patients to involve' - we've moved from people to patients. Needs to be consistent throughout. Also add in something about why you might want to consider having more than one person ie a) public can support each other b) helps redress the power balance in the room and c) the public can not always make a meeting and so, if you have more than one person, it reduced the likelihood that the public voice will be absent at any given time.

p11 Replace the sentence 'Aim to find patients who represent the demogaphics etc' with 'Consider the demographic of those affected etc'. Some readers will take the first sentence to the extreme and it may become a barrier to involving people. I would also suggest losing the sentence 'It can be challenging to access 'typical' members etc' - I'm not sure what is meant by 'typical' here. Need to give some consideration here to the issues of 'representativeness' - when you have only one or two people involved in your research it is unlikely that can be 'the' voice of everyone but they can be 'a' voice. If you want something more representative then surveys etc might be a more appropriate answer.

p11 The authors say that 'the more confident and financially secure are more likely to volunteer'. Need to add in something about researchers have struggled to access certain groups. The sentence 'A lack of resources' - not sure that 'less privileged' is a phrase I would use . Perhaps 'less well off' or something similar?

p11 Need to be careful with the sentence 'Yet this is important..' - some ethnic minority groups might be offended that you are asserting that they tend' to experience lower health status' . Perhaps 'some groups tend to experience etc'

p12 'Patients can contribute to three key functions etc'. Lose this sentence. I'm not sure that it's true. For example they can also be included in data collection (also applies to the penultimate bullet point in 'key messages' on page 15).

p12 'AT an early stage a researcher is advised to discuss roles and task etc' - I would also add in here behaviours or responsibilities.

p13 'INVOLVE has a Patient-Led-Research-Hub to support patients who want to puruse their own research.' Are you referring to INVODirect? This is more a list of organisations who support active PPI in research.

p13 'Researchers wanting to study migraine etc'. Add on to the end of the sentence 'this entails.' (I read it at first as the patients doing aspects of the governance issues).

p15 Either lose the final bullet point on the key messages - I found this confusing - or add in something like 'to ensure you get the public view'.

Is the topic of the opinion article discussed accurately in the context of the current literature?
Yes

Are all factual statements correct and adequately supported by citations?
Yes

Are arguments sufficiently supported by evidence from the published literature?
Yes

Are the conclusions drawn balanced and justified on the basis of the presented arguments?
Yes

Competing Interests: No competing interests were disclosed.

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Author Response 25 Jun 2018

Pat Hoddinott, Dr, UK

Thank you Gary for these very helpful suggestions, in particular the sentences where we could confuse the reader.
Pat Hoddinott

Competing Interests: No competing interests were disclosed.

Discuss this Article

Version 1

Reader Comment 18 Jun 2018

Miles Sibley, The Patient Experience Library, UK

Great paper. In Box 1, under UK Resources, your readers might like to know about the Patient Experience Library - www.patientlibrary.net Thanks!

Competing Interests: No competing interests were disclosed.