Collaboration of patient research partners and scientists improves the outcome of health research

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Structure of this article/content

Introduction

When researchers in the medical sector submit a grant application, they are encouraged to conduct patient involvement in their research. Patient involvement in scientific health research aims to ensure that progress occurs. Patients become involved as patient representatives to ensure that research outcomes better meet the needs of current and future patients. Patient involvement and inclusion can enhance the implementation of the results. Maarten de Wit (2016) writes: "Direct involvement of stakeholders contributes to the enrichment of research agendas and ensures that research results better meet the needs and wishes of end-users. Patient involvement increases the legitimacy of scientific research". Furthermore, to co-produce, there is a need for constructive, open, transparent dialogue between the two sides. NIHR in the UK describes in their co-production in action number three the following key principles for successful co-production:

- sharing of power – the research is jointly owned, and people work together to achieve a joint understanding.
- including all perspectives and skills – making sure the research team includes all those who can contribute.
- respecting and valuing the knowledge of all those working together on the research – everyone is of equal importance.
- reciprocity – everybody benefits from working together.
- building and maintaining relationships – and emphasis on relationships is key to sharing power.

This article outlines different possible methods of patient involvement and demonstrates how they contribute to the relevance and quality of health research. After reading this article, we hope that researchers will better understand patient involvement and patient inclusion in research and how it could meaningfully impact scientific health research to create more alignment between clinical research outcomes and patient -reported outcomes. Patient involvement is becoming an integral part for most industry sponsors and increasingly more research funders too, to demonstrate impact.

This article aims to help researchers consider which kind of patient involvement activities are desirable, and it also provides some guidelines and frameworks to help researchers estimate the budget and time required for patient involvement and patient inclusion. This article can help researchers and patient representatives better identify their roles. Meaningful patient involvement needs good communication between the researcher and patient participant. Patient engagement, patient involvement, or public involvement are terms that can be used interchangeably in different settings and different countries. As a rule of thumb, the latter is about engaging the broader public in understanding science, while patient involvement and engagement are seeing patients play a key and critical role in the design of the research study in different stages. A handy practical guide of these terms can be seen in the EUPATI Glossary Toolbox. EUPATI is the European Patients' Academy on Therapeutic Innovation see here: https://toolbox.eupati.eu/glossary/. In this article, we use the word patient involvement.

Patient involvement in health research

Patient involvement is not simply about informing and involving the study participants in the actual research itself as part of the recruitment, but in patient participation patient research partners are seen as equal partners that have a role to play in the identification of endpoints, in the design of the study protocol, in the dissemination and exploitation of the results and the governance of the actual project in making sure that the research study takes into account what matters to patients. From the viewpoint of the patients: "nothing about us, without us." Summarizing: involvement in research means research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them.

Collaboration with a patient association is one way to undertake patient involvement. There are health research initiatives that form focus groups for a specific study, undertake surveys and a range of both qualitative and quantitative methods and there are also researchers who have extensive networks of patients around them over the years. The advantage of the patient association representatives is that they have access to patient communities. The benefit of local groups may be more engagement and possibly leverage grassroots communities more efficiently.

Rationale for patient involvement in health research

There are various rationales for patient involvement:

1. Patient involvement is an integral part of healthcare research. It is more likely that the study will meet the needs of patients by involving experienced patient experts. Project results will be applied earlier and with more success. (ZonMw, 2018)
2. Patient involvement increases the quality of the research. That is perhaps the most important reason for every researcher. Brett et al. (2012) wrote about this: "Patient and public involvement (PPI) helped gain deeper and more personal insights because of the report users had with participants…", "PPI helped ensure emerging themes and trends were Interpreted from the user perspective as well as the academic researcher perspective, assisted in Identifying relevant knowledge gaps, and final research reports benefitted from being grounded in user experiences", p641.
3. Patient involvement is inspiring, and the researcher has more contact with the (future) patients of research results. Alternatively: "Perhaps the most important benefit of an active stakeholder engagement program is its potential to move research off bookshelves and into practice. If bi-directional relationships sustain over time, stakeholders could serve as ambassadors for high-

integrity evidence where the findings are contrary to generally accepted beliefs. The purpose of patient-centred outcomes research (PCOR) and comparative effectiveness research (CER) is to assist patients, providers, and others in making informed decisions. To accomplish this, the researcher must start to engage the full range of stakeholders in all internships or research" (Concannon, 2012, p 990).

4. Patient involvement from the start, so from the drafting of the research proposal, increases the ethical acceptance of the research. Staley and Elliot (2017) wrote the following about this: "Public involvement in the design stage of research has the potential to increase the ethical acceptability of the research, and to facilitate the decision-making process for research ethics committees", p13.
5. In our society, the participation of citizens and clients is becoming more regular and is a consequence of the complex networks that make up our society. Greenhalgh, Jackson, Shaw, and Janamian wrote in 2016: "*The growing popularity of co-creation in some circles in recent years should be seen as part of a wider change in the science-society relationship. The simple, one-way, and the readily auditable relationship between a group of scientists that undertake research and a foundation that commissions and then uses such research has given way to complex networks or inter-sectoral collaborations and interdependencies and to an ongoing debate about what should be researched by whom, and how*", p407.
6. The patient's voice has increased in health care, and with that, the influence of the patient. The World Health Organization (WHO) stated in 2008 in their world health report that community participation is important in the improvement of healthcare delivery and health equity.
7. Patient involvement contributes to empowering patients. The WHO states that by 2020 patient empowerment is the primary goal that it wants to achieve.

Patient involvement in the different research phases

Every study has the same overall structure. Patient involvement serves a different purpose in every phase of the research and will have different designs at each of these stages.

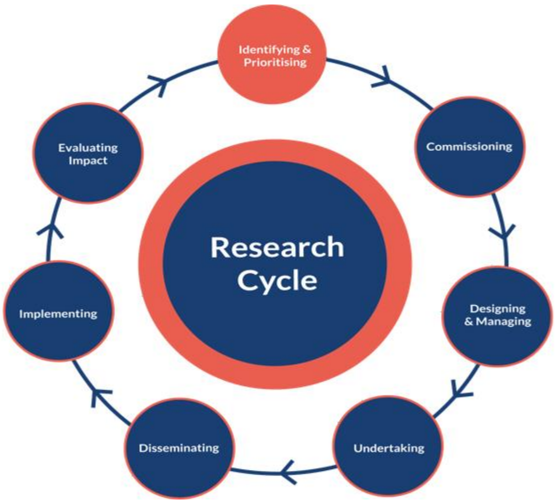


Figure 1: PPI involvement in each stage of the Research Cycle, NIHR

Identifying and Prioritizing

Patient involvement in the preparatory phase of the research includes choosing a research topic based on the wishes and needs of current and future patients. Consultation with patients and representatives in this phase can ensure that a researcher gets a broader view of the research topic from a user perspective. Furthermore, patient representatives provide their unique knowledge, which is experiential of what it feels like to live with a particular disease and to participate in a study as a research participant. Through conversation and dialogue, one gets different perspectives. An expert can look at a problem from a scientific viewpoint but often can miss critical aspects to the end – user. A researcher may have blind spots or tunnel vision, and a patient may have dogmatic views.

By discussing possible topics for further research together, the researcher's agenda is placed next to the wishes and needs of patients so that both parties can decide jointly on research priorities. De Weger, Van Vooren, Luijkx, Baan, and Drewes, 2018, state that the early involvement of the (future) patient or experienced patient research partners makes the process's value better. Early involvement increases the quality of the outcome. Staley and Elliot,2017, confirm that although researchers recognize the benefit of patient involvement, there is less recognition of the valuable contribution of patient involvement in the conceptual, practical, and ethical aspects of the research design. It is slowly changing across Europe as the European Commission has a familiarity and awareness of the importance of meaningful patient involvement and inclusion. It has also become a critical component for most pharmaceutical industries to have patient engagement embedded in different activities in recent years.

If patient representatives are involved in the research design at the earliest stages of the cycle, they can also help with the feasibility of the study. They can advise about inclusion and exclusion criteria, the burden for the study participants, and capacity issues of them. They can help prevent dropout and identify and predict issues with patient recruitment.

Research conduct and operations

Patient representatives can help to find study participants. He/she may have access to a network of potential study participants. Collaboration with a patient representative may generate extra confidence for potential study participants as it has been shown how valuable peer- to- peer networks can be.

An essential part of any clinical research is to inform the study participants properly. Therefore, the study participants need a patient information leaflet with the researched treatment(s) benefits and risks communicated in easy-to-understand language to make informed choices. This leaflet needs to be accessible, written in plain language, and easy to understand for all study participants, not just required by any sponsor or funder.

Therefore, it is wise to involve patient representatives to help preparing the written information for the study participants of any actual study.

During the progress of the investigation it is essential to pay attention to personal needs, manage communication and expectations to ensure that study participants do not drop out during the study. Getting feedback about the participants' experiences can be helping to evaluate any study and enable both researchers and patient representatives to work together in collaboration. Providing study participants', the opportunity to be kept informed of the study's results make it more likely to ensure behaviour adherence with any treatment/protocol.

After data collection it is the researcher's responsibility to record the substantiated results based on the data, but a patient representative may add a different perspective to the data and help sort them according to patient relevance.

Dissemination

After the research has been completed and the conclusions have been drawn, researchers must disseminate the research results to colleagues, doctors, (future) patients, and the public. Peer scientific and clinical networks need to be informed, and results of the findings need to be disseminated at conferences, with the publication of articles in professional academic journals.

Writing scientific articles and giving presentations at (international) congresses is primarily the researchers' task. However, a story by an experienced patient can add a different flavour, provide much needed user perspectives and help attract media attention.

Study participants and patient representatives can play a valuable role here. Staley and Elliot, 2007 say this: "Active involvement of patient representatives in dissemination also helps provide customized information to the public and thereby improves communication " (p. 17).

Implementation

The new treatment must be included in the guidelines during the phase of implementation. Moreover, other actions for implementation should be planned. Most of this work must be done by professionals, but patient representatives can help to accomplish this by sharing his/her experience.

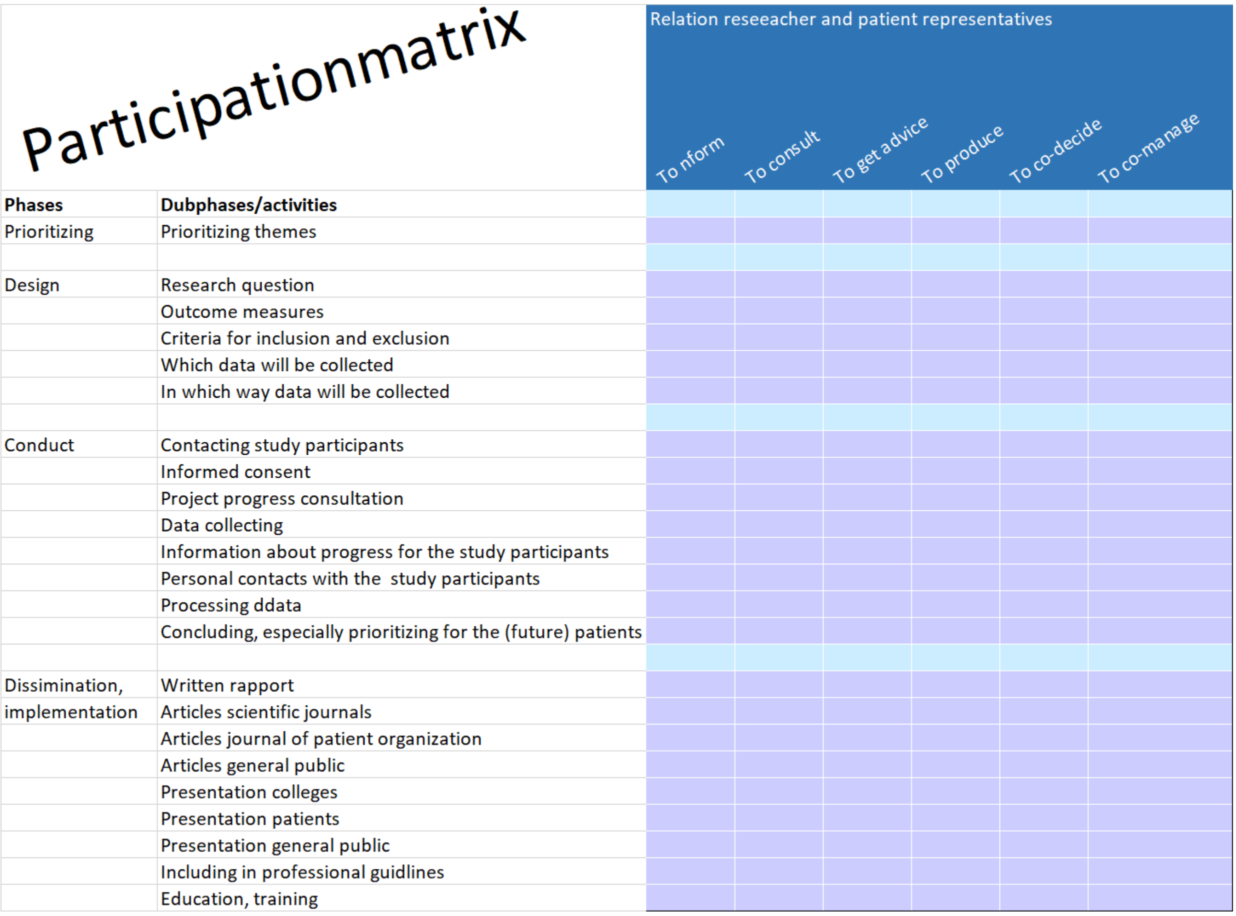
The participation matrix: which role researcher and the patient representatives fill at each stage of the project

Above, we have outlined various tasks for study participants and patient representatives during the research.

Patient representatives have the most impact if they participate at the right time in the correct way in the research project. By 'right,' we mean the moment and method that best suits citizens concerned, with the right levels of knowledge and appropriate time investment included in this matrix.

For example, Smits et al., 2020 developed a conversation tool through which researchers and patient representatives could discuss and explain their involvement in a research project. This tool was formalized and visualized as a 'matrix.' The so-called Involvement Matrix describes five actions (e.g., to inform, to consult, to get advice, to produce, to co-decide, to co-manage), and three phases (e.g., Preparation, Execution, and Implementation) and includes a user's guideline. The phases he used are a little bit different from the phases of the UK NIHR.

The participation matrix is not describing the ideal form of participation. It is a tool for researchers and patient representatives to discuss the roles, tasks and, responsibilities in the different phases of the research.



Participationmatrix

Relation reseeacher and patient representatives

Phases	Dubphases/activities	To nform	To consult	To get advice	To produce	To co-decide	To co-manage
Prioritizing	Prioritizing themes						
Design	Research question						
	Outcome measures						
	Criteria for inclusion and exclusion						
	Which data will be collected						
	In which way data will be collected						
Conduct	Contacting study participants						
	Informed consent						
	Project progress consultation						
	Data collecting						
	Information about progress for the study participants						
	Personal contacts with the study participants						
	Processing ddata						
	Concluding, especially prioritizing for the (future) patients						
Dissimination,	Written rapport						
implementation	Articles scientific journals						
	Articles journal of patient organization						
	Articles general public						
	Presentation colleges						
	Presentation patients						
	Presentation general public						
	Including in professional guidlines						
	Education, training						

Figure 2: Participation matrix based on Smits et al. (2020).

The authors of this paper see this matrix as a tool to visualize and discuss the role and stages of the patient research partners in scientific research but are aware of its limitations. We suggest using this tool before the project starts. The matrix is also very suitable as an evaluation tool.

Critical comment

Are there any disadvantages to patient involvement? Collaboration in the context of an investigation requires a degree of formality (Emonts & Moser, 2014; Moser, et al., 2018). Collaboration requires professional conduct from the researcher but also the patient representative. The endorsement of a project application by a patient association must be a well-considered decision. It is becoming increasingly common practice in many countries to include formal letters of support with signatures from patient organisations and stakeholders in the submission of documentation, often a degree of a legally binding agreement between a patient organisation and research institution is needed on behalf of the board. Of whom? That is why a formal letter with a signature on behalf of the entire board is highly recommended.

In an ideal scenario, not one single individual acts in a representative capacity on behalf of all patients. It can prove to be risky if that individual becomes unwell and cannot continue with the project or takes on the representation of all patients, even if this individual is the chair of an organisation. There is a need for a far-reaching consultation not just with one or two individuals, but to have in place a formal contract between the researcher and the board of the patient association or the group of experiential experts from a focus group of an institution to meet governance standards and to ensure and manage expectations of all parties involved.

A much better way is to have a board member of a patient organisation sit in the project group and enable a consultation that can take place perhaps twice a year between the researcher and the entire association board. To summarize, we recommend at least two patient representatives in health research projects, and where possible, encourage the creation of agreements between different parties.

Some non-scientific final words

After reading this article, we hope the reader has a good idea of what meaningful patient involvement entails and can begin to recognize the benefits of collaboration between patient representatives and researchers in health research.

The authors of this article are not scientists. Therefore, we end this paper with some reflections. The skills, knowledge, and attitudes of the researcher and the patient representatives are, in our view, essential for the success of patient involvement. A good researcher is open to the input of patient representatives and is genuinely curious about their contribution. A patient-participant is willing and capable to share his/her experiential knowledge and lived experience. Is the researcher willing to articulate his knowledge and experience simply so that it is understandable? Does the researcher want to genuinely strive for the best possible result while optimally utilizing the patient representatives? Has the researcher the ability to articulate his scientific knowledge in plain language that can impact on non-scientific audiences? Is the researcher able to integrate what a patient representative puts forward as an essential outcome he/she would want to see in the research? Can the researcher genuinely understand and align where there are divergent views to reach a joint position? Can the researcher dig deeper and understand the user perspective to hear the patient's point and read between the lines on more subtle nuances? If not, can he/she bring in someone that can get that level of trust and rapport needed to ensure patient centricity? Can the researcher plan an investigation so that new input from patient representatives does not lead to unacceptable delays? Does the researcher have the attitude to put his/her contribution in perspective to be open to information from a completely different perspective, namely that of the patient?

We are convinced that the quality of health research improves when researchers and patient representatives collaborate openly and honestly. We welcome the participation and inclusion of patient reviewers in the ERA-Net NEURON initiative.

Glossary

- **Study participants** are persons who participate in health, scientific studies as a patient, who can be patients, potential patients, carers, and people who use health and social care services, and people from specific communities and organisations that represent people who use services. People with lived experience of one or more health conditions, whether they are current patients or not. In this article, we use the word patient involvement. Similar terms used frequently are patient engagement or public involvement (e.g., NIHR or EUPATI).
- **Patient representatives** are patients who collaborate in the research team. Their main responsibility is to give input from the patient's perspective. An important characteristic is that they are not in a dependent position as patients with regard to the researchers; they are completely independent of the researchers.
- **Patient reviewers** evaluate research proposals from the patient's perspective. They can report their findings to the scientific peer review panel, which evaluates the scientific quality. For ERA-Net NEURON, we use the term patient reviewer. A patient reviewer can be a person with lived experience, a patient, a carer of that patient, or a general lay member of the public interested in public health research. A patient review panel pays attention, among other things, to the relevance of the study for (future) patients, the feasibility of the study from the patient's perspective, and in which activities and roles the patient research partners play in the different stages of the research itself. In different countries, for example, the UK, Ireland, the Scandinavian countries, and the Netherlands, and European organisations such as EUPATI and ERA-Net NEURON patient representatives increasingly participate as full partners in the reviewer boards.

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