

Recommendations for Successful Patient Involvement in Scientific Research

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Introduction

The Netherlands is one of the front runners in promoting patient participation in scientific research. This has resulted in several studies focusing on the facilitators, barriers and instruments for meaningful involvement of patients. Patient organisations, as well as researchers and health research funders expressed the need to translate this knowledge into recommendations for daily use. Therefore, PGOsupport, a Dutch networking organisation that facilitates patient support groups to share information and expertise, initiated a series of three meetings, a literature scan and an electronic survey. The objective of the meetings was to exchange knowledge and experience and to develop a list of practical recommendations based on available evidence and expert opinions. In total, 30 representatives from 24 renowned organisations participated in this process. Consensus was reached on a set of 9 recommendations, which are presented and explained in this article.

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Note:

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1. Phases

Patient involvement should be ensured in various phases of research projects, starting preferably as early as possible.

Patient representatives can contribute significantly to each phase of the research cycle (Fig. 1). Studies have demonstrated that patient priorities are not identical to those of investigators and medical professionals. [1] That is why many associations for patients and people with disabilities ('PG organisations') and health funds have made an effort to enrich research agendas with themes that are relevant from a patient perspective.

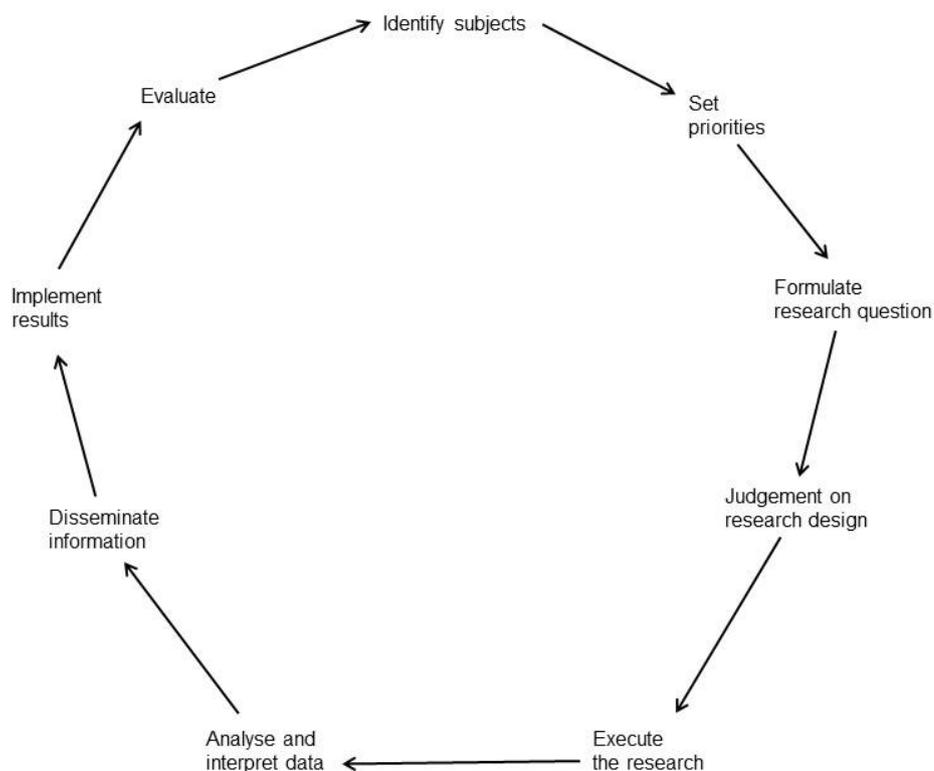


Figure 1. Empirical research cycle.

Patients can also help think through other research phases, e.g. drafting grant proposals and formulating research questions, ensuring questions and themes are included that may be outside of the investigator's scope. For this reason, the Dutch Cystic Fibrosis Foundation started a [Research feedback group](#). [2]

Involving patients in the design phase of clinical research frequently leads to different choices of outcome measures or valuable suggestions for recruiting respondents. Patient contributions often focus on relevance and/or user-friendliness of measuring instruments. [3-5]

The (limited) descriptions of patient representative involvement in data collection, analysis, interpretation and implementation of results, show that also the research process benefits from the involvement of research partners in these phases. [6-8] Finally, patients and their organisations can play a significant role in the implementation and dissemination of study results. [9]

Informal patient involvement scenarios may help to increase awareness of disease impact and have investigators experience the added value of patient participation. Several patient associations say they spend much time and energy building and maintaining good relations with relevant research groups on the basis of equality. An example of this is De Hart&Vaatgroep (for people with cardiovascular diseases) with their [‘College van Ervaringsdeskundigen’](#) (expert cardiovascular patient panel).

2. Patient Role

Integrating patient perspectives in research projects requires a combination of participation methods, on various steps of the participation ladder.

Patients participate in research in many ways according to the ‘participation ladder (Fig.2), e.g. as member of a patient panel, feedback group, guideline working group [10], steering group or scientific advisory board [11]. Sometimes they act as an advisor or research partner [7].

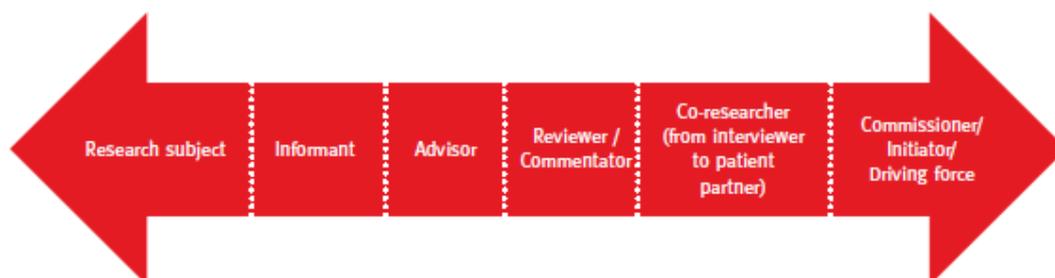


Figure 2. Diversity in the rolls patients can fulfil in the context of scientific research.

In rare diseases, it is often the patient associations that initiate scientific research. [12] We know from patient group experience that patient contributions from different roles are complementary. One or two patient representatives in a research team can help think through the design and conduct of a study, but are unable to represent *the* perspective of the entire target group. A questionnaire can provide this broader perspective. Multiple forms of participation are needed to achieve representativeness and diversity. [13]

An organisation where patients participate in scientific research in various roles is the Dutch Breast Cancer Association (BVN). A Patient Advisory Group, formed in collaboration with the Breast Cancer Research Group (BOOG), advises investigators on patient preferences in an early research phase. For quick member consultations, BVN also uses B-force, a wide circle of highly motivated, hands-on patient experts who can be consulted using an advanced website functionality. Both forms of participation reinforce each other.

3. Recruitment and Selection

Recruitment of patient representatives is supported by a clear job description (profile) and a recruitment strategy focusing on diversity and representativeness.

Most organisations of patients and people with a disability express they (would like to) select on the basis of predefined criteria such as motivation and attitude, communication skills or knowledge of scientific research. It should be noted that different forms of participation require different competencies and skills of volunteers. Consulted PG organisations express the need for an instrument to guide them in selecting the right volunteers for specific tasks.

Alternatively, the initiative for recruitment may lie with the investigator, [14] who approaches suitable candidates through medical professionals. The medical professionals assess patient competencies and the research setting for which the patient is invited. Both strategies involve a risk of bias.

A special form of patient involvement in scientific research is involvement that transcends a single disease. For instance, the Netherlands Organisation for Health Research and Development (ZonMw) and Patient Federation NPCF are currently gaining experience with a broad panel of patient representatives in assessing the relevance of projects for specific programmes (Appropriate use of medication, Efficiency Studies and the Open Programme).

4. Support

Patient representatives should receive individualised information and coaching, both by a good introduction at the start as well as during the research itself.

Depending on individual needs, patient representatives should be supported by the investigators with whom they work together. Both the literature and practice show that the investigator's attitude is crucial. [15][6] The investigator should ensure an open and safe atmosphere during meetings where patients feel free to speak up. This can be achieved by asking open questions and explicitly inviting patients to share their opinion (and/or that of their constituencies). [16] Understandable language and terminology are crucial here. Researchers need to pay attention to the risk of overloading the patient representative. Support can also be given by a patient association, e.g. in the form of information and networking. If the patient role requires patients to contribute not only their own experience, an inventory of their constituency's wishes and needs (in writing) may be a useful resource for patient representatives. [17]

Support starts with aligning mutual expectations of the process and project. This includes providing up-to-date, relevant information. Agreements on a clear job description clarify the role, expectations and time investment of both the patient representative and the investigator. A volunteer contract may help formalise issues as competency, responsibilities, confidentiality, conflict of interest, available support and expense reimbursement.

Sometimes one person is designated as the patient representative contact within a research team. Several PG organisations have assigned coordinators responsible for recruitment, training and support of patient representatives and contacts with (external) investigators. [One of the volunteers](#) took on this role in the Dutch Parkinson Association. Part of his job is to match patient representatives with research projects. The literature shows that it is wise to have at least two patient representatives participating in a research project. This safeguards continuity in the event of disease or drop-out and provides opportunities for prior consultations. [18]

A (generous) expense reimbursement (recommendation 5), training facilities (recommendation 6) and acknowledgements (recommendation 9) may also be regarded as support.

5. Funding

Patient participation costs money and should be budgeted realistically. Patient representative expenses should be reimbursed.

Patient participation costs money and should be budgeted realistically in research project proposals. Adequate budget estimates explicitly include direct expenses for patient representatives and compensation for the patient association related to costs for recruitment, training and support.

De Hart&Vaatgroep reimburses travel expenses when volunteers attend a meeting or training for their work. Some institutions, including ZonMw, earmark funds for patient representatives in the form of attendance fees. In some research projects, patient representatives, in their role as research partner, received a salary.

Agreement on reasonable compensation also requires an individualised approach. The nature of the work, research partners' preferences, and duration and intensity of required contribution are relevant determinants.

6. Training

Training of patient representatives and investigators will benefit the scientific research. Prior to each project with patient participants, the need for training should be reviewed with those involved.

Many initiatives in the Netherlands and Europe [19] to reinforce patient perspectives in research include some form of training of patient representatives. Eurordis, the European umbrella organisation for rare diseases, organises an annual summer school, and EUPATI, the European Patients' Academy on Therapeutic Innovations, offers an intensive training programme and a [toolbox](#) for patient experts. Depending on the initiative and initiator, the focus may be on (biomedical) knowledge of the disease in question, methodology of scientific research, or the value and meaning of hands-on expertise in scientific research. Increasingly, the '[criteriawaaier](#)' (criteria fan) by Truus Teunissen [20] is used as a guide to define criteria from a patient perspective. Other topics for training include discussion skills; research protocol structure; dealing with hierarchy; statistics; and the development process of medicines.

Training can be prepared and carried out by PG organisations themselves, or by external parties, such as Tools Foundation. [PGOsupport](#) offers courses and skills training for various audiences and participation formats. [21] PG organisation volunteers can participate in PGOsupport training free of charge.

Training programs for investigators are only sparsely available and deserve more attention. It is incorrect to assume that investigators have the knowledge and skills required for participatory research by nature. [22]

7. Evaluation

Patient representative contributions to the project and collaboration with investigators should be regularly evaluated.

Studies demonstrated that it is effective to discuss mutual expectations on contributions and the level of participation prior to a research project. [7][23] This will help to manage expectations with respect to time investment, frequency of meetings and tasks involving patient representatives and the level of participation. Importantly, both parties should participate in this discussion on the basis of equality and clearly specify their limits and possibilities. Because we know from experience that both investigators and patient representatives will grow into a project, and needs and expectations may evolve, it is wise to (re-)evaluate the collaboration at regular intervals.

The patient associations ReumaZorg Nederland and the Psoriasis Vereniging Nederland have developed a checklist for the first meeting between a patient representative and a researcher. Evaluative research proved that both parties very much value a clear instrument to meet expectations.

8. Visibility

Examples of patient involvement and the specifics of contributions of patient representatives should be described precisely. The manner of publication should be the subject of discussion between investigators and patient representatives.

Continued and consistent reporting of patient involvement in research and its impact is vital. [46] This includes both positive and negative outcomes.

The Participatiekompas.nl ('Participation Compass') website offers examples (in Dutch) from practice and experience and methods and tools in the field of patient participation. Participatiekompas.nl was developed in 2014 with grants from ZonMw/VSB Fonds and PGOsupport. Many organisations contributed to its development. All stakeholders in patient involvement in research, policies and quality of healthcare may add and find know-how and experiences on the website. [24]

When investigators are encouraged to specifically describe how they will integrate patient perspectives in their project, pseudo participation may be avoided. Also, instruments and methods are better evaluated and developed when their practical implementation is carefully documented. Furthermore, the availability of best practices and learning experiences may encourage other investigators to integrate patient involvement also in their research. Finally, financiers of health research need systematic reporting on the added value of patient participation to legitimise their policies in this field.

Careful publishing about patient involvement is not the same as publishing *at all times*. There may be reasons not to include a (detailed) description of patient contributions in some research publications. First and foremost, the manner of publicising should be the subject of discussion between the investigator and patient representatives, preferably from the start of the project. Patient representatives have an important responsibility to share knowledge, act as co-author, or encourage investigators to pay more attention to this. The anonymity of patient representatives should be ensured at all times, unless otherwise agreed.

9. Recognition

Patient representatives' contribution should be recognised appropriately both during and after a research project.

Although many patients are satisfied with their role as patient representative in a research project, they regularly report receiving little feedback on their contributions to the research process. This could mean that scientific committees overlook to inform the patient panel on projects that have been approved. Or do not give feedback on how the committee

incorporated the assessment of the patient panel in its final decision. Additionally, patient representatives are usually appreciative when they are informed of research outcomes, also when their contributions were completed earlier. Adequate feedback increases motivation for involvement in the future.

There are many ways in which investigators can express their appreciation. In addition to the previously mentioned financial compensation in the form of attendance fees, another way is to make scientific information accessible (library, Pubmed, a subscription etc.), facilitate conference or symposium attendance, or include their names in scientific and non-scientific publications, whether or not as a co-author.

Conclusion

The following associations were involved in the drafting and endorsement of these recommendations. They explicitly approved the inclusion of their names in this publication:

- Borstkanker Vereniging Nederland
- Center for Translational Molecular Medicine
- Crohn en Colitis Ulcerosa Vereniging Nederland
- De Hart&Vaatgroep
- Diabetes Vereniging Nederland
- Hartstichting
- Longfonds
- LevenmetKanker-beweging
- Nationale Vereniging ReumaZorg Nederland
- Nederlandse Brandwonden Stichting
- Nederlandse Cystic Fibrosis Stichting
- Nederlandse Vereniging van Hemofiliepatiënten
- Nederlandse Vereniging van Sjögren Patiënten
- Nierpatiënten Vereniging Nederland
- Parkinson Vereniging
- Patiëntenfederatie NPCF
- PGOsupport
- Psoriasis Vereniging Nederland
- Schildklier Organisatie Nederland
- Spierziekten Nederland
- Stichting Tools
- Vereniging van Mensen met Brandwonden
- ZonMw

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