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ORIGINAL ARTICLE

Whose perspective is it anyway? Dilemmas of patient involvement in the development of a randomized clinical trial – a qualitative study

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ABSTRACT

Background: Patient and public involvement (PPI) is increasingly becoming a requirement in the effort to improve the relevance and quality of healthcare research. We examined how involving patients with lower education levels affected PPI in the development of the MyHealth randomized clinical trial of breast cancer follow-up from the perspectives of the patients and professionals.

Material and methods: Eight women who had completed breast cancer treatment, four with fewer than 10 years of education, were recruited as members of a patient panel advising researchers in the development of the trial. We carried out individual and focus group interviews with panel members and recruiting nurses between April and September 2016. Researcher observations and changes made based on panel feedback were also documented. Patients were asked to evaluate the process according to a PPI theoretical framework with four dimensions: (i) ways of involvement, (ii) research vs. patient concerns, (iii) strength of the patient's voice, and (iv) degree of change. A combination of inductive and deductive thematic analysis was conducted whereby emerging themes were organized using the above framework.

Results: All patient contributors reported high satisfaction with being involved and PPI improved trial materials and recruitment strategy. However, contradictory perspectives between lay and expert approaches to research led to dilemmas not related to educational background. Patients were often more concerned with unmet needs after cancer than with research, and the scientific hierarchy made it difficult for researchers to include the patient perspective if it challenged research requirements. Nurses also faced ethical dilemmas when recruiting patients as PPI contributors.

Conclusions: Our findings challenged the assumption that PPI automatically leads to a broad range of patient perspectives that can directly improve research relevance and quality. This highlights the need for more research and better guidance on the use of PPI in research.

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Introduction

Patient and public involvement (PPI) is becoming increasingly important and required by funding agencies and academic journals [1]. Defined as 'research being carried out with or by members of the public rather than to, about or for them' [2,3], PPI can be traced back to developments in the healthcare sector in the United Kingdom (UK) during the 1990s, where reforms began casting patients as consumers with the right to be heard [4–6]. As a result, patients now play roles such as sitting on grant committees and helping researchers develop studies [3,7].

Besides the democratic rationale regarding the representation of the patient's voice in knowledge production, the underlying assumption of PPI is that nonprofessionals, such as patients, bring unique perspectives that researchers can use to improve the quality of the research project, even though this may be hard to define [7–10]. There is some evidence that PPI can provide knowledge that may be used to develop interventions addressing needs that are more relevant to patients, improve recruitment strategies, and produce user-friendly information and questionnaires [3,7,11]. Other intrinsic benefits of PPI include patients feeling empowered and valued, while researchers report deeper insight into their research area [1,12].

Recent efforts have been made to produce guidelines on PPI for clinical trials [13–16]. Available advice for researchers, however, is still limited and mainly focuses on general principles, such as identifying who should be involved and what contributions are needed [10,17,18]. Furthermore, PPI can be time-consuming and costly for both patients and researchers, which has been found to affect the level of PPI in clinical

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trials [1,3,19,20]. As a result, patients that are involved may tend to be patients who are easiest to recruit, whether these are patients from organizations already established for involvement purposes or motivated patients who volunteer for the job [8].

It may be hypothesized that such patients who actively participate in PPI often have higher levels of education and health-related knowledge. This selectivity raises the concern of a potential 'professionalization' of these patients and whether they provide perspectives that may be representative of the general patient population [8]. Accordingly, INVOLVE, the UK national authority on PPI, advocates the practice of 'inclusive' involvement where patients from seldom-heard populations are given the opportunity to influence the research process [21]. The aim is to ensure that PPI in research does not end up leading to the development of healthcare interventions that are tailored only to those with strong voices, thus marginalizing 'the very groups the system was intended to involve' [5]. To our knowledge, however, no study has examined inclusive involvement in the development of a clinical trial.

This current study is nested in the MyHealth randomized controlled trial comparing a nurse-led breast cancer follow-up program with standard oncologist-led care in women who have completed primary cancer treatment [22]. The trial protocol incorporated the establishment of a panel of patients who act as advisors to researchers, which is one of the most common forms of PPI in research [20]. As MyHealth aimed to be a personalized intervention, tailored to the individual needs of breast cancer survivors, it was important that especially cancer patients from less advantageous backgrounds were represented on the patient panel, so that their opinions regarding the intervention and its materials could also be heard. This provided the unique opportunity to examine inclusive involvement, defined in this trial as the involvement of patients who also had lower levels of education, in the development of a clinical trial.

Theoretical framework

Successful PPI in healthcare research involves the dynamic interaction of different forms of knowledge, notably that of the patient and the researcher [23]. In order to conceptualize and understand these interactions, we drew on the four-dimensional 'cube model' (Figure 1), which was developed by Gibson et al. [5] based on the social and philosophical works of Arnstein [24], Habermas [25], Bourdieu [26] and Fraser [27] regarding the role of power and inequality in participation and decision-making. Adapted to the research context, the cube model proposes understanding PPI along three main dimensions and a fourth overarching one: (1) *ways of involvement*, which refers to the plurality of ways in which patients were engaged; (2) *research concerns versus patient concerns*, which refers to the interface between research and patient priorities; (3) *strength of voice*, which refers to how much influence patients had on the decisions made; and finally, (4) *degree of change*, which summarizes the extent the research project was able to accommodate and change based on patient involvement [5,23].

The original aim of this paper was to examine how involving patients with lower levels of education affected PPI in the development of a clinical trial from the perspectives of the patients, recruiting nurses and researchers involved. However, during the study, contradictory perspectives between lay and expert approaches to research led to dilemmas not related to educational background. In this paper, we report on these findings using the four dimensions of the PPI framework above.

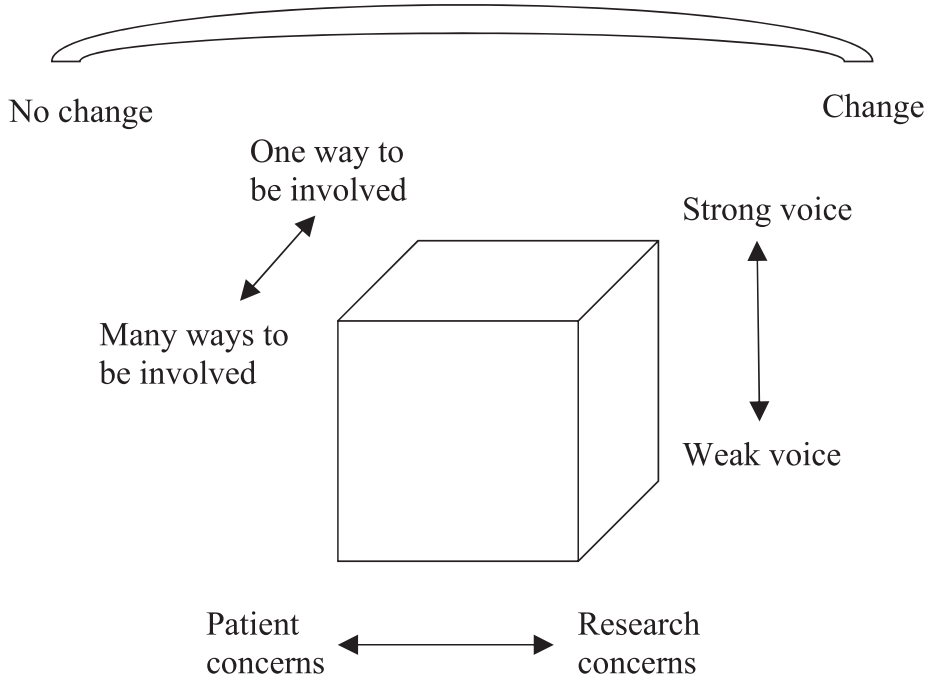

Figure 1. Four dimensions of conceptualizing patient and public involvement in healthcare research based on the cube model (Gibson, 2017). Reproduced with permission from the author.

Material and methods

Design

This was a multiperspective qualitative study nested in the development of the above-mentioned MyHealth trial. A qualitative method was selected because it allows the analysis of subjective experiences and recognizes the researcher's role as an explicit part of the knowledge-creation process [28]. In the MyHealth intervention group, fixed follow-up visits with an oncologist are replaced by patient education sessions with a trained nurse, followed by systematically collected patient reported outcome measures (PROMs) over three years, which alert nurses if treatment is required [22]. The nurse sessions and PROMs target physical and psychological symptoms after breast cancer and are described in detail elsewhere [29].

Using the PPI guidelines mentioned above [13,17,18], trial researchers identified which patients to involve, the areas requiring patient input, recruitment procedures and decided on the use of the focus group interview as the preferred method of gathering a range of patient perspectives through facilitated discussion [30]. For the purpose of this study, individual interviews were carried out a few weeks before and after the focus group meeting to collect data on patient motivations, expectations and evaluation of the process. All interviews were carried out based on a semi-structured interview guide and were audio recorded. The key topics covered by the interview guide for the pre-interview, focus group interview and post-interview are presented in Box 1. Approval for the MyHealth study, including the patient involvement plan, was granted by the Danish Capital Region's Health Research Ethics Committee (No.: H-16035885) and by the Danish Data Protection Agency (J.no.: 2016-41-4728).

Participants

A panel of eight breast cancer patients was established, which has been suggested to be an optimal number for focus group interviews [30]. The panel was established such that half of the members consisted of women whose highest educational level was up to 10 years of basic school. Patients had to meet MyHealth inclusion criteria (i.e. female, completed primary treatment for breast cancer with no clinical sign of recurrence and be able to read, understand and speak Danish) and were currently attending follow-up appointments at the Department of Oncology at Zealand University Hospital, Naestved, Denmark. From 21 April to 8 June 2016, two recruiting nurses from the department's clinical research unit, LD and EMR, screened 802 patients using hospital records and identified 228 who met both the criteria for MyHealth and an inclusion list created to ensure an even mix of patients based on age ($\leq$60 years, $>$60 years), civil status (married, single) and stage of follow-up ($\leq$1 year, $>$1 year).

Nurses approached eligible patients after their scheduled follow-up appointment with the oncologist. They briefly explained the project and the rationale of involving patients from a range of educational levels before asking about the patient's educational background. Education was chosen as a screening variable because it is relatively easy to ask about compared to income, and is associated with a range of relevant indicators of socioeconomic status including income and health literacy [31,32]. Eligible and interested patients received a pamphlet that outlined the role of the panel and the time commitment required. Due to conflicting nurse schedules, patients who did not turn up for their appointments and patients who left early before the nurses could approach them, a limited number of patients could be invited each day. Thirty-one patients were invited before the eight consenting patients were obtained. The main reason given for declining was lack of energy and resources. One member later withdrew before the first interview citing the inability to handle the involvement process, leaving seven women in the panel. Characteristics of the panel members are shown in Table 1. None had ever been involved in research in any way. Only three members had fewer than 10 years of education.

Data collection

Data were collected between April and September 2016 and consisted of individual and focus group interviews with

Box 1. Key topics covered by the interview guide for the pre-interview, focus group interview and post-interview with members of the patient panel.

Pre-interview	Focus group	Post-interview
• Introduce the MyHealth project and clarify expectations regarding the role of the patient contributor • Elicit motivation for participating and expectations for involvement • Elicit their healthcare experiences during and after cancer treatment • Profile patient's education and health literacy levels	• Patient perspectives on the overall intervention, including any concerns about being followed-up by a nurse instead of a specialist • Revision of materials (informational brochures for patients and partners, educational materials on symptoms and symptom management and outcome questionnaires) for ease of understanding and clarity • Optimize recruitment procedures and explanation of the randomization process to trial participants • Presentation and discussion of the IT platform used to collect patient-reported outcome questionnaires	• Debrief contributor on changes made based on contributor feedback • Explore patient contributor experience and perspectives on the involvement process • Carry out evaluation of the involvement process based on the "cube model" of PPI

Table 1. Characteristics of patient contributors.

	MH01	MH02	MH03	MH04	MH05	MH06	MH07
Age (years)	76	64	67	71	52	54	56
Stage of follow-up	>1 year	>1 year	>1 year	≤1 year	≤1 year	≤1 year	>1 year
Married	Yes	Yes	Yes	No	No	Yes	No
Education	≤10 yrs	>10 yrs	≤10 yrs	>10 yrs	>10 yrs	≤10 yrs	>10 yrs
Employment status	Retired	In employment	Retired	Retired	In employment	In employment	In employment

Table 2. Key themes from data analysis in relation to the four dimensions of the cube model.

Themes and issues	Dimensions of the model
• Acceptability of written materials and interviews as method of involvement	1. Ways of involvement
• Discrepancy between researcher expectations and patient motivations for PPI	2. Patient concerns versus research concerns
• Researcher-perceived mismatch between efforts and results	
• Dilemma of relevant versus "irrelevant" knowledge within the research framework	3. Strength of patient's voice
• Patient acceptance of "the way things are"	
• Change to trial only possible within confines of research requirements	4. Degree of change
• Recruitment and ethical challenges	

panel members, an interview with the recruiting nurses and researcher documentation of the process. The focus group interview was held on 24 June 2016, took place at the local Danish Cancer Society's counseling center and lasted three hours. Panel members were provided beforehand with the materials requiring PPI input: MyHealth information brochures for patients and their partners, educational materials on symptoms and symptom management, and patient reported outcomes questionnaires. All seven panel members took part in the preinterviews and postinterviews but due to conflicting schedules only three were able to attend the focus group (MH01, MH02, MH03). For the remaining four women, the topics for the focus group were discussed in the postinterview.

In the interview postfocus group, panel members were asked to evaluate the involvement process using a form with a figure illustrating the cube model. JAS carried out all the personal interviews, BLH, JAS and LS facilitated the focus group interview together with a representative from the company developing the electronic platform to be used for collecting questionnaire data in MyHealth, while BLH interviewed the two recruiting nurses at the hospital. Nurses were interviewed regarding the screening and recruitment process. Researcher documentation consisted of a nurse log of the recruitment process, reflections and notes by JAS and BLH after interviews and coding sessions, and a log over the changes made in response to panel feedback. While JAS only played a role in this current study, BLH and LS were also attached to the MyHealth trial as PhD students.

Data analysis

All the interviews were digitally recorded and transcribed verbatim. We carried out deductive or "theoretical" thematic analysis as described by Braun and Clark [33] using the four dimensions of the cube model to explore our research question, '*How did educational level affect patient involvement in research?*' However, as we also remained open to issues of potential interest in the data itself, the analysis evolved into a more inductive approach early in the process, as other themes began emerging that did not necessarily relate to the patient's educational background. BLH and JAS read all the interview transcripts and coded the data independently, after which identified themes were verified and refined in a collaborative process among BLH, JAS, TTS and PEB. Data management and analysis were carried out using NVivo 11 for Windows [34].

Results

We organized our findings within the four dimensions of the cube model (*Ways of involvement, Research concerns versus patient concerns, Strength of patient's voice and Degree of change*) and summarized the key themes in Table 2. One theme, recruitment and ethical challenges, did not fit any dimension and is reported separately.

Ways of involvement

Gibson et al. developed this dimension based on Bourdieu's idea of social capital, which confers higher status to certain groups in society (e.g., doctors) compared to others (e.g., patients) [26]. Since different social groups produce different forms of knowledge, that is, patients will express themselves in different ways, effort is required to diversify the ways in which patient knowledge is elicited [5]. In this study, panel members were involved though individual and focus group interviews and had to go through quite a bit of written materials. In a pre-interview, one panel member said:

> *I have to say, if I have to read a whole bunch of papers… I'm telling you, I CAN'T do it. I won't do it! (MH01, 76 years)*

Thus, using written material as the sole mechanism of involving patients can be a limitation for certain patients, such as MH01, who cited having bad eyesight, was our oldest panel member and had 9 years of formal schooling. In such situations, verbal and face-to-face modes of involvement may help. MH01 did, however, review all the materials for the focus group interview and here, all panel members openly discussed their opinions and even helped each other understand aspects of the intervention.

It appeared that regardless of their educational background, the women in this panel felt comfortable engaging

through verbal expression. In the evaluation, all the women agreed that the experience was positive:

> *It was great to hear the experience of others and meet other women like me. I hope I was able to help … it's an interesting project.* (MH02, 64 years)

Research concerns versus patient concerns

This dimension refers to the balance between research and patient priorities during the involvement process [5]. We found a large discrepancy between the researcher assumption that patients who participated were motivated by a desire to contribute to research and actual patient motivations, which were often based on a specific concern or unmet need. This was illustrated by MH03 and MH01:

> *That's why I joined this … I feel my doctor (GP), he wants to push it away … if I even hint at having any pain, he'll say they have to take care of it at the oncology department. And at the oncology department, they say I have to go to my GP … so I'm really stuck in the middle.* (MH03)

> *Well, I don't know if there is a doctor in your project … I'm not sure this has anything to do with the project … but I actually want to ask, what are the chances really of getting cancer again if I stop taking these (anti-hormone) pills? Because I have to admit, I think I'm going to stop.* (MH01)

Several of the patients turned out to be suffering from pain, hot flushes and other side-effects of adjuvant endocrine treatment. As the involvement pamphlet listed the doctors who were involved in the project, it is possible that some patients saw the opportunity to get a second opinion. In fact, during the focus group where LS, an oncologist, was present, patients indeed asked her about several treatment-related issues. In fact, only one patient, MH04, a retired physiotherapist with 40 years of working experience in a healthcare setting, specifically mentioned research as a motivation:

> *I would like to contribute, if I can, to research, because it is incredibly important.* (MH04, 71 years)

Interestingly, JAS, had the most difficulty keeping to the interview guide with this panel member, because she was highly interested in talking specifically about research on diet and cancer. In fact, panel members often had to be refocused on giving feedback on MyHealth because they got caught up talking about their illness and experiences instead. We noted:

> *There seems to be a mismatch between our effort and results … we had to really work hard to get the relevant knowledge out of the contributors. On the other hand, we received a-lot of input and information … but they were outside of the topics in our interview guides.* (JAS and BLH, notes from coding session)

It seems that unknowingly, researchers were relating to the patients mostly through the lens of research and evaluating their contributions accordingly – patient contributions were deemed relevant only when they fell in with research priorities. This is a paradox since the researchers were involving patients with the aim of focusing on patient concerns.

The strength of the patient's voice

The third dimension of the cube model refers to how much influence patients had on the decisions made [5]. We expected that patients with higher levels of education would contribute with a 'stronger voice,' that is, be more critical, thus having more influence. As mentioned above, we found that panel members regardless of their background, were willing to share their opinions. However, it was only in cases where patient contributions could directly be used in the predefined areas of the trial, that the panel members directly influenced the decisions made.

For example, in MyHealth, researchers wanted to ensure that the randomization process was explained clearly to potential participants to reduce the risk of drop-out among those who feel cheated when allocated to the control group. During the focus group, panel members discussed this issue with LS, who would be the recruiting doctor for the trial, and concrete sentences on how to explain the randomization process and the equal importance of the intervention and control groups were developed and written into the recruitment plan. Similarly, MyHealth brochures and patient education materials were revised to make it easier to understand based on the feedback received from panel members, and patient concerns regarding answering questionnaires electronically were noted down. In these straightforward examples, patient voice might be considered strong, regardless of patient education. In fact, patients who had the most difficulty understanding matters were the ones who contributed the most in helping researchers clarify procedures and informational materials.

However, in the case of outcome questionnaires, patient voice carried no influence. Many of the panel members expressed difficulty in answering some parts of the questionnaire, citing answer categories that were difficult to judge and specific items that they were unsure of. This posed a dilemma for the researchers, who agreed that some of the wordings in the questionnaire could be confusing, even though the selected scales were validated and widely used. However, replacing validated scales would reduce the quality of the research according to the norms of quantitative research. Thus, despite strongly voiced opinions from the patient panel, the research team did not make any changes to the outcome measures on the questionnaire.

Interestingly, when this was explained to panel members during the debriefing part of post-interviews, everyone easily accepted it as 'the way things are.' Furthermore, in their evaluation of this dimension, all the panel members reported feeling that their voices were heard but expressed uncertainty over whether their contributions were helpful:

> *In any case, I've expressed what I think (about the project) but whether or not it's helpful, I don't know, that's for you to decide … but I have in any case given my opinion.* (MH05, 52 years)

> *I really feel like you listened … I'm not the sharpest in class but I think, well, it's also good that you get feedback from us on the ground … what do* we *think? But whether or not you can use it (our input) for anything, I don't know!* (MH01)

Thus, patients perceived being involved in research as a positive experience but did not feel that they were in a position to judge whether their contributions had an effect on the final decisions made.

Degree of change

This fourth overarching dimension of the cube model summarizes the extent the research project was able to accommodate and change based on PPI [5]. Patient feedback led to changes and improvements in recruitment strategy, brochures and educational material, the electronic platform created to collect questionnaire data, as well as helping researchers ensure that questionnaire items were generally understandable and not offensive. However, changes were not made to item wordings or answer categories in order to uphold the validation of the scales. In this aspect, the interests of research were privileged above those of the patients and we return to this in the discussion.

Recruitment and ethical challenges

This final theme concerns the difficulty in explaining the relatively new concept of PPI in research to patients and the ethical considerations involved in recruiting seldom-heard patients. Traditionally, for a patient, being involved in research has meant being involved as a subject or study participant, not as a collaborator or advisor. The recruiting nurses explained that a lot of care had to be taken to clearly explain what was expected of the patient and what PPI in research is, finding it helpful to use the pamphlet developed to explain the role of the patient panel and began explaining it as a 'pre-research project' to help develop a 'future research project.' Although this effort helped the panel members understand their role as contributors in the development of the MyHealth materials, some of the panel members still thought that they would be participants in the MyHealth trial and asked in the post-interview, '*When will I start* (the intervention)?'

The nurses also brought up ethical considerations regarding the selection of potentially vulnerable patients.

> *I had a patient who had bad hearing … I had to shout to tell her about the project. And she said, yes, she would really like to join. But as we talked, I asked her about how she was doing. And actually, she wasn't doing well at all. Her partner had just died, suddenly, and she was facing a lot of legal and practical issues that needed to be solved. I mean, she was an elderly lady, mid-70's, in grief and facing all these problems … in the end, I said to her, You know what? You should not join this. I won't tell you more about this study. You need to talk to your doctor.* (EMR)

This highlights the dilemma between the democratic aspect of inclusive involvement, where the goal is to ensure that seldom-heard patients are also represented, and the ethical aspect of what it means to involve these patients, some of whom may be too vulnerable physically or psychologically. The implicit evaluation of patients carried out by the recruiting nurses could also be seen as a breach of study protocol if the patient is willing to participate in the project. The nurses, however had a different perspective:

> *You risk getting patients who say yes, out of the goodness of their hearts, but who do not have the resources … either they drop out in the long run, or they continue and ruin themselves. As project nurses, it's something we take seriously … it is our most noble task to guard and take care of our patients.* (LD, project nurse)

Generally, however, the approached patients expressed interest in being on the patient panel and the nurses reported that many with more than 10 years of education were disappointed when told that they could not be included because only spots for those with lower educational levels remained to be filled.

Discussion

To our knowledge, this is the first study to investigate inclusive involvement in research. Informed by the four-dimensional PPI theoretical model that maps the interaction between lay and expert knowledge [5,23], we wished to examine how involving patients with lower education levels affected PPI in the development of a clinical trial from the perspectives of the patients, recruiting nurses and researchers. We found that the idea of inclusive involvement met strong support from the nurses and approached patients. In line with previous findings [1,3,7], we also found that patient contributions helped improve various aspects of the intervention, such as improving recruitment strategies and producing user-friendly information.

Despite allocation of adequate resources, receptive attitudes from all parties, and efforts to follow PPI guidance, such as identifying and planning the 'who, what and how' of involvement [13,14,35], researchers still experienced several dilemmas, mainly arising from the contradictory perspectives between lay and expert approaches to research. For example, there was a mismatch between the vast experiential knowledge that was being produced by the patients and what could be implemented by the researchers. This might not have been the case if resources were available for involving patients earlier in the process, for example, in determining intervention aspects in the protocol stage, since many of the concerns that were 'irrelevant' were issues already targeted by the intervention, such as information regarding the side effects of treatment.

The decision to use psychometrically robust scales that were actually sub-optimal from the patient's point of view highlights the difficulty researchers can face trying to reconcile patient perspectives with research criteria based on the scientific hierarchy of evidence, which positions objective methods above subjective opinions [36]. This may lead to 'tokenism,' that is, PPI that is merely symbolic and carries no influence [37]. Thus, new hierarchies may need to be developed in order to accommodate other types of research and alternative ways of measuring outcomes that prioritizes the patient's voice [38,39]. However, as we found, this is no easy task and more research needs to be done in exploring new ways of aligning scientific considerations and what matters to patients [40]. It is important to underline that this research

should be based on scientific knowledge and not on opinion, and that scientific criteria, for example, from qualitative research, should ensure the scientific framework of the patient perspective in research [41].

A related dilemma was the gap between effort and results. Substantial expenses were incurred by planning, recruiting and interviewing the patient panel based on the assumption that by giving patients the opportunity to contribute, researchers will directly be able to apply the lay knowledge obtained to improve the research project. However, it is hard to evaluate how the changes that ended up being made in the project actually improved research quality. Although there is increasing documentation of the impact of PPI on research [7,42] and the UK Medical Research Council has developed the public involvement impact assessment framework (PiiAF) [35], evidence of improved research quality due to PPI is still sparse, possibly because this is hard to define due differences in patient and researcher norms. A tool for evaluating the quality of PPI from the patient's perspective has recently been developed [43] but whether perceived quality of PPI among patients is associated with increased quality in pre-identified areas of impact remains to be studied.

Finally, the ethical dilemma faced by the nurses highlights a paradox that the very patients needed to ensure that PPI represents the perspectives of marginalized groups may be the ones who are not able to bear the work required. This dilemma between excluding patients who are deemed "not able" to contribute on the one hand and potentially affecting research outcomes on the other warrants further attention because it parallels a more general problem in PPI concerning the decision of which patients to involve. It is not uncommon for patients to receive some research training, for example, learning scientific terms and methods, to help them contribute in the most optimal way [8,44]. However, in its extreme, this may result in the creation of 'professionalized' lay experts, who no longer represent the general patient's perspective but rather a research perspective, which defeats the purpose of PPI.

This study has several limitations. The involvement of patients relatively late in the development of the MyHealth trial and the use of semi-structured interview guides with predefined areas may have limited how researchers could use patient input. Also, several of the researchers involved in collecting and analyzing data for this article were also involved developing in the trial, which may bias interpretation of the data. The strengths of this study include the possibility of examining the involvement of an entire panel of seven women instead of just one or two patient representatives, the fact that these women were systematically recruited to ensure representation of lower education levels and the multiple interviews carried out to capture patient perspectives before, during and after the involvement process.

Our results highlight the complexities involved in integrating the patient perspective in the research process. Successful patient involvement involves the dynamic interaction of patient and researcher knowledge, but this gives rise to many dilemmas. More specific guidance needs to be developed in collaboration with funders, researchers and patients, which includes how to manage the tensions between patient and expert priorities in specific research settings.

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