Author’s response to reviews

Title: Recruiting patients as partners in health research: A qualitative descriptive study

Authors:

Lidewij Vat (eva.vat@med.mun.ca)
Devonne Ryan (devonne.ryan@mun.ca)
Holly Etchegary (Holly.Etchegary@med.mun.ca)

Version: 1 Date: 07 Aug 2017

Author’s response to reviews:

August 7, 2017

RE: Response to reviews of manuscript RIAE-D-17-00009

Recruiting patients as partners in health research: A qualitative interview study, by Lidewij Eva Vat, MSc; Devonne Ryan, MSc; Holly Etchegary, PhD

Dear editors,

Thank you for the opportunity to revise and resubmit our manuscript; we truly appreciate reviewers’ time and careful attention to the paper. They have raised some legitimate and important points and concerns, and we have incorporated nearly all of their suggestions. We used red text in the revised paper for easy identification of changes; please find our detailed point-by-point response below.

Reviewer reports:

Reviewer #1:

Major comments

* The authors describe this study as filling a key gap in the literature, although they do not reference other seemingly related studies and systematic reviews. The article could benefit from reference to the broader literature in this area, and further explanation as to what their study adds to this body of literature. See for example:
Thank you for this suggestion. We had cited several reviews, including Domecq and colleagues, but were unaware of the review within the area of rare diseases. We have slightly re-worked the introduction to better explain what the current study adds (lines 85-88). We very much liked the rare diseases review paper, and have incorporated it into the discussion (lines 350-351, 370-371). Again, thank you for pointing it out.

* The paper could benefit from a definition of both "recruitment infrastructures" (or ways to find each other? in the plain language summary) and "recruitment strategies", as well a description of the relationship between these themes. Can each strategy be used within each infrastructure, for example?

Thank you for sharing this suggestion. We agree that the difference between strategies and infrastructure was not clearly highlighted. We have incorporated the definitions (lines 102 - 105) and we better explained the different infrastructures. Each strategy can be used within each infrastructure, however the recruitment process and approach might be slightly different. In the third-party model and directory model patients are often recruited based on their interest in becoming involved in health research rather than an interest in a specific research project. Third-party models and directory models require also a recruitment strategy to encourage patients to sign up for the registry. This can be done via various methods such as social media, partnering with advocacy groups, via health care professionals, community events etc.

* Along the same lines, can the authors comment on the differences between "partnering recruitment" as a recruitment strategy (line 235) and the "directory model" as a recruitment infrastructure. To me, these seem quite similar and it again raises the question as to what the difference is, if any, between a recruitment infrastructure and a recruitment strategy.

Good questions. We understand that these can seen as similar. In our opinion the third-party and the directory model is slightly different as this is a structure or platform specially created/available for patient engagement in health research. We defined recruitment strategies as the methods participants used to find patients who would be interested in becoming a patient partner. Recruitment infrastructures were defined as the structures or platforms available to facilitate recruitment. We also better defined ‘partnering recruitment’ [lines 223-226]. This includes recruitment in collaboration with an organization or group who have members or represent a particular patient perspective such as advocacy groups or charitable organizations. Another partnering strategy could be partnering with a marketing company who have panel
members. The difference with the third-party or directory model is that the members are not specifically already recruited for engagement in health research.

* In the title and abstract, the study is referred to as a "qualitative interview study", but this terminology does not describe a study design but instead a method of data collection. I think it would be more appropriate to refer to this study as a qualitative descriptive study. See Sandelowski’s 2000 paper

This is an excellent point. We are aware of Sandelowski’s paper and more recent update (2010). We agree and have changed the title and abstract accordingly.

* On line 125, the authors describe using qualitative description to summarize the data. Qualitative description is a study design and not a data analysis technique. Most commonly in qualitative description studies content analysis is used. I would suggest naming the study design earlier on in the methods section (and the title and abstract in place of "qualitative interview study") and reworking the analysis section to describe the techniques of content analysis.

Thank you for reminding us of this important distinction. Upon re-reading, we agree that a slight re-working of the data analysis section was in order. We introduced the ‘Study design’ section (line 114), and moved the explanation of qualitative description study designs to this section. As we had explained the content analysis methods in the data analysis section, we now more correctly identify it as content analysis and not qualitative description as a data analysis technique.

* Face validity does not appear to be the right term to describe sharing the results of the thematic analysis with the Patient Advisory Council (Lines 97 and 142). Face validity refers to whether something appears to be measuring what is intended to measure, as in a psychometric analysis. Instead, I wonder if the authors used this strategy of sharing results with the Patient Advisory Council as a form of peer debriefing, or peer review?

Also an excellent point. We also struggled somewhat with how to correctly describe this. In the main, we intended for the patient advisory council to comment on whether these findings made sense from their perspective and whether these seemed reasonable recruitment strategies, as well as whether any were missing from their perspective. But we agree face validity is likely not the best phrase for describing this. We thank the reviewer for suggesting peer review, and we have slightly revised our wording to reflect it (lines 152, 155-160)
* It is my feeling that because the Patient Advisory Council identified a range of issues not covered in the interviews, it seems as if data saturation was not obtained. Can the authors comment on the concept of data saturation, sample size, and the ability to draw conclusions from this study when data saturation does not seem to be obtained?

This is a really important point, our thanks for raising it. We had noted the small sample size and the obvious limits this raises for generalizability (line 394). We have added a section about data saturation to the limitations section (lines 397-399). It’s an interesting question – whether data saturation was obtained, or whether in fact, patients have different ideas about recruiting patient partners than do researchers and so different themes would emerge from the data (we suspect the latter). In our interviews, very few new themes or ideas were raised after the first 8-9 interviews, so we are reasonably confident data saturation was reached in this particular data set. However, it is very possible that research explicitly comparing patient and researcher perspectives on recruitment would reveal some differences. We now note this in the discussion (lines 400-402).

* Patients’ perspectives seem important to obtain, as patient partners ultimately should be consulted for activities that impact them. It would likewise be interesting to compare patient partners’ perspectives with researcher perspectives in terms of recruitment strategies. One patient partner interview was conducted, but this seems inadequate to explore this perspective appropriately. Given the research goals differ (i.e. a researcher vs. patient partner perspective) it would seem that either more interviews with patient partners are needed to more comprehensively explore this perspective, or it should be left out altogether since it is inadequately explored. Can the authors comment as to why one interview is sufficient?

We couldn’t agree with the reviewer more! Of course the patient perspective is equally important, and as the review by our patient advisory council suggests, may well reveal different concerns and perspectives. By way of providing some context for the reviewer – we chose to focus on researchers and patient engagement leads for this project as we had a very short time frame and funding (~4 months; funding for research assistant support and transcription only). We felt we could not fully explore both perspectives within that short time frame and funding environment. Yet we felt strongly that this kind of work needed to at least start, motivated in large part by our involvement on the national working group. This group often discussed what recruitment strategies they were seeing in their own provinces, and all the leads in that group commonly talked about fielding questions from local researchers about how to recruit patients as partners. So we felt a good place to start, even with our limited time and funding, was to begin to systematically collect data from those currently recruiting patients as partners (the leads and researchers they identified). But we truly struggled with not including a patient perspective at all, and that is why we eventually decided to invite an interviewee from Patients Canada. We agree
with the reviewer that this perspective certainly cannot adequately nor fully explore the patient perspective, but we thought at least a national perspective from a patient organization would be a useful addition. In the end, however, the reviewer is absolutely correct that future research explicitly exploring and comparing these two perspectives is needed, and we have added that to the discussion (lines 408-409).

* In "the exception" section, it seems that this respondent did not understand the intention of this research project, and I therefore question the credibility of these data. It is also interesting to read this person's comments and consider that many of the same limitations that apply to recruiting research participants may also apply to recruiting research partners. Can the authors comment as to how the misconception of this particular respondent, as raised again in lines 343-344, was addressed during the interview? If the interviewee did not understand the intention of the study and line of questioning, it is curious that the interviewer did not correct this individual during the conduct of the actual interview.

In many respects, the reviewer is correct that recruiting participants versus recruiting research partners is very similar, and perhaps this is a notable finding itself. This is always a tricky situation for interviewers. On the one hand, you do indeed want participants to understand the purpose of the study, but a certain amount of objectivity is also required. We did not want to prime participants too much, nor expressly direct their answers. The study information letter and consent form were very clear about the purpose of the study, and this researcher was recruited through snowball sampling from another participant as a researcher with experience in partnering with patients in research. Thus, it may well be the case that this participant does not see a distinction between participants and research partners. Nonetheless, we felt it was important to be honest about this viewpoint. It could have easily been left out or ignored, but this is inappropriate in considerations of rigor in qualitative research. In fact, we think this perspective is very important as it reminds us that even in this sample – those purportedly with some experience in recruiting patients as research partners – misunderstanding still exists in the distinction between patients are participants vs. partners. It is our hope that even in some small way, this study can help raise awareness of that important distinction.

Minor comments

* It may help to clarify the purpose of the study throughout as to being about describing strategies that have been used to recruit patients, as opposed to simply "documenting strategies" in the abstract, plain language summary and main text. In the plain language summary, for example, the authors state "The goal of this study was to look at ways of recruiting patients as partners in health research" but it would be more clear and reflective of
what was done to report something like "The goal of this study was to describe ways that patients have been recruited by researchers and patient engagement leads".

* The plain language summary could benefit from a definition of 'patient partners'

Thank you for raising these points. We have replaced the sentence in the plain language summary as recommended, and replaced ‘documented’ with ‘described’ in the abstract and other parts of the main text when noticed. We also added an explanation of patient partners in the plain language summary.

Reviewer #2: This is generally a clearly written and structured paper. There are some aspects of terminology which I would suggest need to be addressed, particularly the use of the word 'engagement' which is generally not used in the UK to mean 'involvement' in research. The authors cite Angela Coulter's paper on patient engagement in support of their opening claims about the benefits of 'engagement', but she was referring to engagement in one's own care and decision-making, not research so much. Similarly INVOLVE was set up not for general public involvement in the NHS but specifically for research involvement. Generally I felt the introduction was a little uncritical about the claims made for the impact of PPI.

We thank the reviewer for pointing this out. We feel the description of INVOLVE is largely accurate (though we have replaced ‘engagement’ with ‘involvement’ to be accurate), but you are absolutely correct about Coulter’s paper, which did indeed refer to patient involvement in their own care and decision making. Much of the beginning work on patient engagement in research is certainly informed by works such as hers, with a focus on patient engagement in care decisions and health systems decision-making more broadly (e.g., Julia Ableson’s work in Canada). However, we did indeed need to make that distinction more clear (lines 62-63). The distinction between engagement and involvement is a tricky one. In our national conversations, terminology continues to be a contentious discussion (with some disliking use of the term patient, let alone engagement or involvement). We felt unable to fully tease apart these distinctions as it was not the purpose of the article. We adopted our country’s definition of partner, engagement and engagement in research to be consistent with national norms, but we take the point that terminology certainly varies among countries and projects (and indeed is one of the challenges to patient engagement in research).

I agree that there has been little research into how people are recruited for PPI. However, I feel the paper is a little thin and descriptive as it stands, and misses some opportunities to take a more critical stance. The sample size is relatively small, and focused almost exclusively on professionals. The inclusion of one patient partner, recruited through a patient advocacy organisation, does not really give us enough feel for alternative perspectives. (Incidentally the
manuscript is inconsistent in parts of the text as to whether the sample was patient engagement leads and researchers or not (see plan English summary v abstract, for example).

These are all worthwhile points. First, we have been more consistent now in the plain language summary and the abstract regarding the participants. Our thanks for pointing that out. We take the reviewer’s point about the descriptive nature of the paper. In truth, the study was designed primarily to be descriptive and give a comprehensive summary of the recruitment methods employed by our participants, as well as a summary of their perceived challenges and facilitators. We feel this is the strength of the paper as (to our knowledge), no other study has focused exclusively on describing recruitment strategies in this context. We did note the small sample size, but have now added lines to the discussion about data saturation and limits on generalizability (beginning line 394). We also take the point about the patient interview. Please see our response to reviewer 1 regarding the patient partner interview. In the main, this is clearly an area for future research (explicit comparison of patient and researcher perspectives).

As it stands, the analysis focuses on what might be described as 'surface' barriers to recruitment such as public awareness, time, skills, compensation, health status, patient motivations - but does not go deeper into questions of relative power between researchers and patients, or researcher behaviour, control and assumptions. I would recommend something like Peter Beresford's Beyond the Usual Suspects report to stimulate thinking on hierarchy and exclusion, and how (whether consciously or not) the system is designed to marginalise some kinds of people. Related to this, the paper does not consider how - once people have expressed an interest or been identified - they are ultimately selected. Is there a requirement for a CV, a formal application form, and interview? Is there a job description with essential criteria? All of these recruitment processes may serve to make recruitment fairer and more transparent - or to exclude and delegitimize patients whose profile doesn't 'fit' what the researcher wants.

These points are all useful and indeed, we did ask our participants for samples of any kind of recruitment documents (such as application forms, advertisements, job role descriptions, and the like). Part of our thinking at the outset of the study was to compile a toolkit of sorts that might include useful templates for others seeking to recruit patient partners for research. Unfortunately, we did not receive enough of these to create a toolkit. Our focus in this study was to describe the kinds of recruitment strategies that participants used to recruit patient partners, not on these sorts of specific details, but the reviewer is correct that these requirements (or lack thereof) could indeed influence the equity of recruitment. Similarly, the study was not designed to explore power differences or how the system is designed to marginalize some people, though we agree this can certainly be the case (and have added some lines to this effect in response to the fourth reviewer’s comment – see below). In the end, the data are descriptive (as they were meant to be), and while this certainly can be criticized for not being particularly ‘deep’ we believe they will be useful to researchers looking for a place to start in their recruitment strategies. Much as reviewer 4 noted, the array of recruitment approaches and their barriers and facilitators are not often
reported, and we sincerely hope this will add value to research teams, particularly those new to patient engagement (and in our jurisdiction, most are!).

The researcher comment on p.9 'I'm not looking to recruit just anybody' is a can of worms waiting to be opened. Several times I thought there were missed opportunities to challenge the surface narrative of what researchers say and critique their stance. The negative case analysis, for example, can be presented as mistaken understanding on the part of the interviewee, or it can be an opportunity to delve into why some researchers make no distinction and what this says about their attitudes and knowledge (or lack of). It felt to me that too often the burden was on changing what patients do, think and know, and not on researchers and their cultural assumptions.

To provide context for the quote on p. 9 – this patient engagement lead was actually working hard to be specific in his recruitment strategies, rather than wasting time and resources compiling a large roster of potential partners and in his words, “not knowing where to connect them”. As noted, his aim was to recruit patients based on the research focus and to directly meaningfully connect them with a project. Rather than having them in a directory and might not being able to connect them to a project. His concern was to identify and recruit patients with lived experience of a very specific condition, as is often the case in the traditional model. This need not be a bad thing, and our patient advisory council also noted the importance of the match between patients and projects (lines 374-375), and other literature has reported that patients are most interested in research specific to their own condition (ref 28). It was certainly never our intention to place burden on patients in what they think, know or do, and we hope our data do not give that impression. We worked hard to consult our patient advisory council before beginning the study, to provide them with the results section of the paper, and to incorporate their comments into our discussion. Please see our response to reviewer 1 regarding the negative case analysis. We hope that by including it, we can help raise awareness of the important distinction between patients as participants vs. patients as partner.

On content of the findings, health system recruitment seemed to be partly a question of recruitment through prior research participation not just being a patient in someone's care. This could be drawn out more.

We agree, our thanks for pointing this out. We have added a line to make this distinction more clear (lines 261-263).

It was also not completely clear to me how the recruitment strategies would mapp onto the infrastructure. Partnering recruitment sounded rather similar to the third party model. The choice of strategy will also depend on the aim of involvement. If the purpose is to expose researchers to lived experience of a particular condition, for example, community outreach is unlikely to be helpful unless the condition is very common.
Please see our response to reviewer 1 regarding the recruitment strategies and infrastructures. We agree that the choice of strategy depends on the aim of involvement and in particular the perspectives and skills needed for the project. We added a section in the discussion [lines 338-344] about which strategy might be helpful for recruiting a certain group or perspective.

Reviewer #4: Thank for the opportunity to review this interesting article.

Some might argue that there is a common sense approach to recruiting patient research partners and guidance already exists (from INVOLVE and others) which recommend similar methods of recruitment to those highlighted in this article. However this research provides valuable insight into the array of recruitment approaches and describes the advantages, disadvantages, barriers and facilitators to each - knowledge which is less well understood and reported. The article also provides a useful framework for others in the field to consider approaches to recruitment.

We are also struck by the common sense approaches related by our participants, and we sincerely thank the reviewer for acknowledging the value in the descriptive data about the array of approaches, with their barriers and facilitators. It is our sincere hope these will be helpful, particularly to researchers new to the area of patient engagement.

The article is well written and structured, and provides a clear account of the qualitative research methods. I therefore recommended publication, subject to a few minor revisions for the authors' consideration:

1. Plain Language Summary - This sections needs to standalone as a clear summary of the whole article. Therefore the following points will improve clarity.

* Clarify what is meant by 'leads' (line 20).

These are all important suggestions. We have added an explanation of ‘leads.’

* Briefly expand on the 'three ways that could help researchers and patients to find each other' (line 21). As a standalone section, this statement leaves the reader wanted to know 'which three ways' (though I appreciate that these are described in depth in the main body of the article and this may have been intentional?).

Certainly we were concerned about length and the amount of detail in the plain language summary, but the reviewer is correct that the ‘three ways’ reference does leave the reader wanting more! We have tried to add more detail, while respecting the length.
* The fourth recruitment strategy, 'Partnering' (line 26) is a little vague and needs a little more explanation.

Agreed. We have added an explanation.

* Three recruitment models listed as 'traditional, third-party and directory' is a bit vague and needs a little more explanation

The additional information provided about the ‘three ways’ noted above should now help clarify these models (explained in more detail in the main text of course).

2. Clarify who conducted the interviews (line 117).

Done. Devonne Ryan, one of the authors. Now line 135

3. Assuming that the author(s) conducted the interviews, as well as designed the study and analysed the data, it might be useful to state the 'researcher(s) stance' and briefly report, as best as possible, how one's preconceptions, beliefs, values, assumptions and position may have come into play during the research process. The authors have provided a negative case example as a measure of balance but I think some researcher reflexivity might be additionally useful.

Word limits often preclude such reflection, but we welcome the opportunity to do so (lines 155-160). We agree it is an important part of rigor in qualitative research, though word limits often preclude the ability to discuss this in any depth. Thank you for pointing it out.

4. Regarding the level of investigator agreement in the data analysis, how high is 'very high'? Please clarify.

Yes of course, agreement was over 90% on key themes (line 151), and even when there was discrepancy, it was usually over the exact wording to use to express the same theme.

5. On this issue of 'health system recruitment, it might be useful to reflect on the power balance between clinicians recruiting their patients in a clinical setting for patient involvement, as
described by Locock et al, The power of symbolic capital in patient and public involvement in health research. Health Expect. doi:10.1111/hex.12519

Many thanks for making us aware of this article. Indeed the notion of power may be particularly relevant for those models of recruitment between clinician-researchers and their patients. We have incorporated the articles to our paper (lines 353-360).

6. I found the statement 'not applicable' to the consent to participate (line 421) a little strange given that this was a interview study and you are wishing to publishing the data.

Now line 468.

Thank you for catching this! We should have included this heading with the ‘ethics approval’ a few lines above it. We mistakenly were thinking of this as consent for publication. We have removed the not applicable, as the reviewer is of course quite correct.

I hope these suggestions are useful for improving what is already a good article.

Again, we are grateful for the opportunity to respond to reviewers and resubmit the paper. Their suggestions were valuable, and we believe incorporating them does make the paper stronger. We look forward to hearing from you, and please do not hesitate to contact us should you require anything further.

Best regards,

Eva Vat and Holly Etchegary

--