

PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	Exploring academics' views on designs, methods, characteristics, and outcomes of inclusive health research with people with intellectual disabilities – A modified Delphi study
AUTHORS	Frankena, Tessa; Naaldenberg, Jenneken; Cardol, Mieke; Meijering, Jurian; Leusink, Geraline; Van SchroyensteinLantman- de Valk, Henny

VERSION 1 - REVIEW

REVIEWER	Ruth Northway University of South Wales, United Kingdom
REVIEW RETURNED	20-Mar-2016

GENERAL COMMENTS	<p>This is an interesting study and overall the paper is well written and clearly structured. I do, however, have some areas of concern that I feel need to be addressed. First the paper seeks to explore the potential of inclusive health research for people with intellectual disabilities. Within such an approach the inclusion of people with intellectual disabilities is fundamental and yet within this study their involvement seems to have been very limited. Furthermore academics / professionals are defined both within the title and body of the paper as the 'experts' which seems to reinforce historical views of people with intellectual disabilities as not being able to actively contribute to research that focuses on their lives and experiences. This is not in anyway to deny the expertise of those who participated in the study: they are experts in their field but I would expect a more inclusive definition of 'expert' in a study regarding inclusive research and people with intellectual disabilities. There thus seems to be a disconnect between the philosophy of the research approach that forms the focus of this study and how this particular study has been undertaken: people with intellectual disabilities have seemingly been marginalised within a study that seeks to explore their inclusion. The authors do note that the Delphi methodology precluded the inclusion of people with intellectual disabilities but perhaps a modified approach would have been possible or, at the very least, there could have been some discussion of the findings of the research with people with intellectual disabilities who have experience of being involved in research and their views could have been reflected as another expert group. If this did not occur (and therefore it is not possible to include it) then I would suggest that there needs to be a more critical discussion of how 'expert' is being used within this study, a stronger rationale provided for not including people with intellectual disabilities, and that this is more fully noted as a key limitation of the study.</p> <p>Whilst inclusive research is not always synonymous with participatory research there is an overlap as acknowledged by the</p>
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	<p>authors. My second area of concern, however, is that the growing body of literature concerning participatory research and people with intellectual disabilities is not used to underpin the discussion in this paper. Within that body of literature there is considerable discussion concerning how such approaches have been used and developed with people with intellectual disabilities, how approaches have been adapted and how power issues have been addressed. Whilst such papers do not always focus on health issues they could, nonetheless, make a methodological contribution. Furthermore the work of authors such as Oliver and Finkelstein regarding issues of power and control within wider disability research could also help to further contextualise the discussion.</p> <p>Overall, therefore, I feel that whilst this paper raises some interesting issues and does further discussion regarding inclusive health research the areas outlined above need to be addressed prior to publication. I hope that these comments are helpful in developing the paper further.</p>
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REVIEWER	Elisabeth Zeilinger, Andreas Kocman University of Vienna, Faculty of Psychology, Austria
REVIEW RETURNED	12-Apr-2016

GENERAL COMMENTS	<p>The paper entitled "Exploring experts' views on designs, methods, characteristics, and outcomes of inclusive health research with people with intellectual disabilities – A modified Delphi study" with Manuscript ID bmjopen-2016-011861 describes an interesting study on inclusive research with people with ID. It is well written and a meaningful contribution to this line of research. Therefore, publication is supported.</p> <p>The following points should be part of a revision before publication:</p> <p>Generally:</p> <ul style="list-style-type: none"> . Please use precise, ideally quantifiable terms and expressions for reporting of data. <p>Abstract:</p> <ul style="list-style-type: none"> . State the number of experts in the abstract. . Why is collaboration italic? <p>Introduction:</p> <ul style="list-style-type: none"> . Not all readers might be familiar with the BMJ patient revolution. In addition to its aim, please describe what kind of collaboration the term describes. <p>Method:</p> <ul style="list-style-type: none"> . Please describe the reason for following up the Delphi process with in depth interviews in more in detail . Please indicate the study that the current research is part of (current publications,...). . Please describe why the co-researcher with ID was not included in the second round and why the results from his first round are not available. It would be very interesting for readers to get to know the opinion of the co-researchers with ID on this study about inclusive research. . You are currently contrasting "people with ID" to "experts" when
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describing the Delphi process. Please consider rephrasing, especially since people with ID were participating as co-researchers.

- . Please provide more information on the expert panel (academic background,...) of the entire sample including non-respondents.
- . Please define the term cognitive interview.
- . Please provide information on how the specific experts were selected for in-depth interviews and provide more information on these experts.
- . Why was agreement only achieved if the median was on one end of the scale? High agreement is also reached if most experts consider an item as not important. Level of (Dis-) agreement should thus be measured as a degree of variance between replies, not the content of these replies.
- . Consider publishing the questionnaires with the study (e.g. as supplement). It would give the reader a better insight into the whole study.
- . In the data analysis section the authors describe three criteria for their definition of agreement regarding a characteristic or outcome. These can be subsumed to only one criteria: more than 75% need to have chosen the response category "very important". The rest is redundant. Please change this section and describe it more comprehensible.
- . Please give examples for disagreements between the two researchers who coded independently, and how these were resolved.

Results

- . Please include more information on dropout rates. I.e. were non-respondents from round 1 excluded from round 2? Were there differences between respondents and non-respondents?
- . Please describe the approach resulting in table 2 more in detail.
- . Provide a clear indication as to how many experts emphasized a certain aspect or replied in a certain way both within the Delphi process or within the interviews. Refrain from using non-numeric quantifiers (many, plural indicating all participants,...)
- . The Delphi method aims at facilitating agreement among a group of persons. Please indicate the general level of agreement among the group after the second (final) Delphi round.
- . Report on interview results and rationale of Delphi findings probably overstates agreement within the group of experts by reporting findings of one homogenous group ("According to them", "they voiced", "Experts envisioned a tool"). If all experts specifically indicated the exact same set of aspects and arguments, then please report so by stating numerals ("all nine experts within the sample stated congruently that..." "Hundred percent of the sample replied that...", "during the interviews all nine experts argued that..."). However if this is not the case, please refrain from indicating statements not provided specifically by all experts (especially in the interviews).
- . Declining agreement might be due to dropout rates. Does this effect persist if only replies from people participating in both rounds are included in the analysis?
- . Adding information on IDs of experts providing certain quotes is helpful. However, no overview on experts characteristics linked to these IDs is provided?

Discussion

- . Please discuss the decline of agreement within the Delphi process. This is unusual and in contradiction to the aim of the Delphi process. Thus more information on possible reasons might be valuable.

VERSION 1 – AUTHOR RESPONSE

Reviewer 1: Ruth Norway

1. We wholeheartedly agree with the reviewer's response on the lack of involvement of people with ID in this study and are aware that the manuscript as such does not reflect our efforts. We have tried to give insight on the actions we have taken to collaborate with people with ID. However, as this paper specifically addresses academic's views, it goes beyond the scope of this paper to elaborate on this as much as we would like. Our collaboration with co-researchers will be a standalone reflection paper, as there is much to learn from the challenges we encountered.
2. Likewise, we agree with the query regarding the terminology used for academic researchers in this study. We have changed "experts" into "academics" throughout the whole manuscript.
3. We thank the reviewer for pointing out the body of literature on participatory research and we have included this in the discussion of this paper. We agree that power issues are of great importance to both inclusive research and participatory research and literature by authors such as Oliver are of great added value.

Reviewer 2: Elisabeth Zeilinger and Andreas Kocman

Generally:

4. We recognise and understand the reviewers' query regarding the usage of statements such as "according to them", "they voiced" and "experts envisioned". In this study, the rationales and interviews are used to identify emergent themes. Academics had the opportunity to address any theme they felt was important, and, therefore, themes were not systematically discussed. As a result, using numerals to indicate the level of agreement will give a biased picture. We have tried to minimise the usage of aforementioned statements throughout the paper. Additionally, in the methodology section we have described how we identified emergent themes.

Abstract:

5. We have stated the number of academics in the abstract.
6. We have rewritten the sentence with "collaboration" in *Italic*, as we believe that presenting the approaches collaboration and control in *Italic* contributes to the readability of the manuscript.

Introduction:

7. We have given further explanation of BMJ's patient revolution.

Method:

8. We have described in more detail the reason for taking on a Delphi methodology followed by in-depth interviews.
9. We have indicated the study that the current research is part of.
10. See query 1 by reviewer 1.
11. See query 2 by reviewer 1.
12. We have provided more information on the expert panel by means of Table 2: Descriptives.
13. We have defined the term cognitive interviews.
14. We have provided more information on how academics have been selected for in-depth interviews. We have added information on the interviewees by means of Table 2: Descriptives.
15. We agree with reviewer 2 that it was not clear why this manuscript only addresses criteria regarding characteristics which were identified by academics as "very important". This manuscript focuses specifically on characteristics and outcomes suitable for inclusive health research, and, therefore, characteristics and outcomes which are scored as "very important" are of main importance. This query made us aware that we have not explicitly stated this in the manuscript. Thus, we have done so by stating we are looking for "the most important characteristics and outcomes" of inclusive health research in the aim of this study.
16. We have published the questionnaire as an online supplement.

17. We agree with reviewer 2 that the agreement criteria we used were unnecessarily elaborate. Firstly, in order to identify the most important characteristics and outcomes, a median of 5 is needed. Secondly, in order to identify whether agreement was reached on the most important characteristics and outcomes an IQR of 0 is needed. Thus, we have now simplified our agreement criteria to aforementioned two. Agreement criterion “more than 75%” is redundant.

18. We have given insight into how the researchers discussed the qualitative analysis by means of an example.

Results:

19. We have included more information on the dropout rates by means of Table 2: Descriptives. In this table, it can be seen that the majority of the academics who dropped out are Health and Disability researchers.

20. We have described the approach resulting in Table 2 (now Table 3) in the methods section under “questionnaire design”, Topic I.

21. See query 4 by reviewer 2.

22. See query 4 by reviewer 2.

23. Hereby we would like to respond to the queries by reviewer 2 regarding the decline of agreement among academics and the dropout rate. We could not find a decline of agreement in our manuscript as such. We agree that the dropout rate is indeed an important methodological issue in Delphi studies and it would be interesting see whether the dropout in this study has an effect on the levels of agreement. However, as this is not a methodological paper, it goes beyond the scope of this research to elaborate on this within this paper. Additionally, to our knowledge, a decline in agreement is not unusual in Delphi studies as it provides experts the opportunity to reach agreement, but does not force them to agree.

24. We have included more information on academics’ IDs by means of Table 2: Descriptives.

25. See query 23 by reviewer 2.

VERSION 2 – REVIEW

REVIEWER	Ruth Northway University of South Wales United Kingdom
REVIEW RETURNED	01-Jun-2016

GENERAL COMMENTS	<p>Thank you for revising and resubmitting this paper. I feel that most of the issues raised during the initial review have been adequately addressed. There are, however, a couple of areas where I feel some slight modification may still be helpful.</p> <p>First in the strengths and limitations section at the beginning of the paper it might be more accurate to change the phrase 'to approach scientifically patients' involvement in...' to 'to approach scientifically from the perspective of academics patients' involvement in...'. This would also require some rephrasing on p16.</p> <p>Whilst you do highlight later in the paper that the views and experiences of colleagues with intellectual disabilities who advised on the project will be explored in another paper it might be helpful to also acknowledge in the introduction that you consider these views to be important and that they will be explored elsewhere.</p> <p>It is good to see that you have drawn upon some further literature concerning participatory research and the use of such an approach with people with intellectual disabilities. I feel, however, that further literature could usefully be drawn upon to strengthen the paper</p>
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	<p>particularly the discussion section. For example you note that many of the guidelines developed in relation to participatory research focus on different client groups and may not, therefore, address the specific needs of people with intellectual disabilities. However, there are a number of studies that have been published in recent years that include people with intellectual disabilities and which provide useful guidance concerning the implications of using such an approach, the adjustments that may need to be made, and the outcomes of such research from the perspective of people with intellectual disabilities. It may be that the word limit for the paper has constrained both your discussion and the use of wider literature. Nonetheless if there is the capacity to include some of this literature in the paper then this would be helpful.</p>
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REVIEWER	Elisabeth Zeilinger, Andreas Kocman Faculty of Psychology, University of Vienna, Austria
REVIEW RETURNED	09-Jun-2016

GENERAL COMMENTS	<p>We appreciate the changes that were made to the manuscript. Our concerns were addressed sufficiently. We regard the paper as a useful contribution to the body of literature. And we especially look forward to the future publication of the reflection paper on the collaboration with co-researches with ID.</p>
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VERSION 2 – AUTHOR RESPONSE

Reviewer 1: Ruth Norway

1. We have adjusted the sentence in the “strengths and limitations” section at the beginning of the paper. Likewise, we have made the same adjustments on page 16.
2. We have emphasised in the Introduction section that people with ID’s perspectives will be studied in another paper.
3. We have added recent literature on participatory research with people with ID in the discussion section.