PEER REVIEW HISTORY

BMJ Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form (**http://bmjopen.bmj.com/site/about/resources/checklist.pdf**) and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

ARTICLE DETAILS

TITLE (PROVISIONAL)	Approaches to Governance of Participant-Led Research: A Qualitative Case Study
AUTHORS	Grant, Azure; Wolf, Gary; Nebeker, Camille

VERSION 1 - REVIEW

REVIEWER	Susan Cox University of British Columbia Canada
REVIEW RETURNED	19-Sep-2018

GENERAL COMMENTS	This is an important and novel contribution to the literature on research ethics and participatory research design. I enjoyed reading it and found the explanation of study goals, design, methods, analysis and findings to be clear and well-organized. I have only a few suggestions for minor revisions: 1) check figures in the flowchart on recruitment -- how can it be that 21 participants completed a project if only 20 received a box of equipment? 2) provide a timeline -- when was the study initiated? how long did each phase take? 3) Link discussion to wider literature in field of participant role in ethics and ethical review. Consider inclusion of other relevant literature on questions of participant conceptions of risk, why people participate in research and importance of trust in researchers (commented on in discussion). See for example: Townsend Anne, Taylor Kim, and Susan M. Cox. (2014). Conceptions of risk regarding a chronic illness survey: Perspectives of participants, researchers, and ethics review board members. IRB: Ethics & Human Research;36 (5):13-20. Cox, Susan M and Michael McDonald. (2013). Ethics is for Human Subjects Too: Participant Perspectives on Responsibility in Health Research. Social Science and Medicine, 98: 224-231. McDonald, Michael, Anne Townsend, Susan M Cox, Darquise Lafrenière, Natasha Damiano Paterson (2008). Trust in Health Research Relationships: Accounts of Human Subjects Journal of Empirical Research on Human Research Ethics, 3 (4): 35–47. Guillemin, Marilys et al. (2018) Do Research Participants Trust Researchers or Their Institution?. Journal of Empirical Research on Human Research Ethics. 13 (3): 285-294 The whole question of participant involvement in identifying relevant ethical issues is a VERY significant one even in more traditional research designs. Participants are seldom asked about

	their experiences of research participation and whether they have insights that investigators could benefit from hearing. This point could be made in the discussion and would enhance the overall relevance of the piece. 4. Check for minor typos and grammatical errors.

REVIEWER	Corine Mouton Dorey Institute of Biomedical Ethics and History of Medicine, University of Zurich, Switzerland
REVIEW RETURNED	23-Oct-2018

GENERAL COMMENTS	It is an interesting and needed paper as PLR, quantified-self and citizen sciences approaches to research are developing fast, and there is a lack of ethical understanding. The case study methodology is appropriate for exploring the issue. We missed the phase two results (submitted elsewhere for publication but not available here). For the phase 3, the results are presented in a quantitative way (%) that is not appropriate to a qualitative case study and needs to be corrected. From what is reported, we understand that participants are aware of the importance of an ethical reflection even for low-risk research, but multiple approaches are necessary to engage in the ethical reflection. Learning process is a major benefit, at all levels. Few quotations are reported: they interestingly point out concepts of trust, group solidarity, responsibility and coercion, which could have let to further reflect on accountability and reciprocity in the discussion. The title "guiding principles for PLR" is not appropriate. We could not follow how these principles have emerged from the analysis of the case study. It is more about "approaches to PLR governance". Then the link between the results and the discussion is not so clear. For instance, the focus of the section on "guiding principles for PLR" is on participant individual requests for: transparent and continuous information on the study, access to expert for understanding the protocol, individual interest for the research question, right to withdraw, benefice and free choice to participate. All these aspects relate to the conditions of an informed consent, which is not clearly mentioned there. The more innovative ethical aspects of "no one-time decision", flexibility, inclusivity could have contributed to introduce the idea of a PLR-governance. PLR-governance could thus have included former findings on reciprocity and trust. Finally the discussion presents only the case of the US human research regulation. The participants came from 5 other countries in Europe. This is a limitation. NB: the references 38 and 39 are inverted.

VERSION 1 – AUTHOR RESPONSE

Reviewer: 1

Reviewer Name: Susan Cox

Institution and Country: University of British Columbia, Canada

Please state any competing interests or state ‘None declared’: None declared

This is an important and novel contribution to the literature on research ethics and participatory research design. I enjoyed reading it and found the explanation of study goals, design, methods, analysis and findings to be clear and well-organized.

Thank you very much, we appreciate that you enjoyed reading the MS and found it valuable!

1.1) I have only a few suggestions for minor revisions: 1) check figures in the flowchart on recruitment -- how can it be that 21 participants completed a project if only 20 received a box of equipment?

Response 1.1: We see how this is confusing. The 4 individuals who were organizers were not shipped a box of supplies as they were already in the office. That means 20 additional people + 4 = 24 individuals started the project. Three sent back their supplies unused, meaning 24-3=21 people completed the project.

1.2) provide a timeline -- when was the study initiated? how long did each phase take?

Response 1.2: We have created a timeline, listed as figure 2, to be inserted just above “Results”.

1.3) Link discussion to wider literature in field of participant role in ethics and ethical review. Consider inclusion of other relevant literature on questions of participant conceptions of risk, why people participate in research and importance of trust in researchers (commented on in discussion).

See for example:

Townsend Anne, Taylor Kim, and Susan M. Cox. (2014). Conceptions of risk regarding a chronic illness survey: Perspectives of participants, researchers, and ethics review board members. IRB: Ethics & Human Research;36 (5):13-20.

Cox, Susan M and Michael McDonald. (2013). Ethics is for Human Subjects Too: Participant Perspectives on Responsibility in Health Research. Social Science and Medicine, 98: 224-231.

McDonald, Michael, Anne Townsend, Susan M Cox, Darquise Lafrenière, Natasha Damiano Paterson (2008). Trust in Health Research Relationships: Accounts of Human Subjects Journal of Empirical Research on Human Research Ethics, 3 (4): 35–47.

Guillemin, Marilys et al. (2018) Do Research Participants Trust Researchers or Their Institution? Journal of Empirical Research on Human Research Ethics. 13 (3): 285-294

Response 1.3: These suggestions are highly relevant and we have added them as citations in our introduction and discussion. We have also expanded our section on participant perception of the risk of coercion

1.4) The whole question of participant involvement in identifying relevant ethical issues is a VERY significant one even in more traditional research designs. Participants are seldom asked about their experiences of research participation and whether they have insights that investigators could benefit from hearing. This point could be made in the discussion and would enhance the overall relevance of the piece.

Response 1.4: We agree that this is an important question that should be both cited and called out explicitly. We have elaborated on this idea in the introduction on page 5. Additionally, we have incorporated this point into the last paragraph of the discussion (added statement is highlighted): “This ethical review in PLR requires a common stake among all participants. This common stake means that all who take part in the project share an investment in the conduct and outcomes of the research. This stake even extends to those in traditional research conditions, in which greater attention to the participant experience stands to benefit both participants and researchers (11).”

1.5) Check for minor typos and grammatical errors.

Response 1.5: Corrected and highlighted in yellow.

Reviewer: 2

Reviewer Name: Corine Mouton Dorey

Institution and Country: Institute of Biomedical Ethics and History of Medicine, University of Zurich, Switzerland

Please state any competing interests or state ‘None declared’: None declared

It is an interesting and needed paper as PLR, quantified-self and citizen sciences approaches to research are developing fast, and there is a lack of ethical understanding. The case study methodology is appropriate for exploring the issue. We missed the phase two results (submitted elsewhere for publication but not available here).

We are very pleased to hear that Corine found this an ‘interesting and needed paper’ – and we hope that once the two papers are published, they can work as a pair!

2.1) For the phase 3, the results are presented in a quantitative way (%) that is not appropriate to a qualitative case study and needs to be corrected. From what is reported, we understand that participants are aware of the importance of an ethical reflection even for low-risk research, but multiple approaches are necessary to engage in the ethical reflection. Learning process is a major benefit, at all levels.

Response 2.1: We have revised the presentation of the results under “group communication” to express the information using examples/quotation rather than percentages. Several quotes have been added and explained under the “Group Communication” (now renamed “Group Communication to Enable Ongoing Ethical Reflection”). Instead of listing percentages, these quotes describe how different forms of communication enabled participants to a) learn material and get their questions answered, b) engage in ethical reflection during experimental planning and revision, c) think about their own perspective by hearing other participants’ thoughts, and d) fit ethical reflection into a busy schedule.

2.2) Few quotations are reported: they interestingly point out concepts of trust, group solidarity, responsibility and coercion, which could have let to further reflect on accountability and reciprocity in the discussion.

Response 2.2: We agree that the MS could benefit from more direct quotation of the participants. As stated above, more exemplary quotations have been added.

2.3) The title "guiding principles for PLR" is not appropriate. We could not follow how these principles have emerged from the analysis of the case study. It is more about "approaches to PLR governance".

Response 2.3: The authors agree, and have changed the title to “Approaches to Governance in Participant-Led Research: A Qualitative Case Study”.

2.4) Then the link between the results and the discussion is not so clear. For instance, the focus of the section on "guiding principles for PLR" is on participant individual requests for: transparent and continuous information on the study, access to expert for understanding the protocol, individual interest for the research question, right to withdraw, benefice and free choice to participate. All these aspects relate to the conditions of an informed consent, which is not clearly mentioned there. The more innovative ethical aspects of "no one-time decision", flexibility, inclusivity could have contributed

to introduce the idea of a PLR-governance. PLR-governance could thus have included former findings on reciprocity and trust.

Response 2.4: We have clarified the link between the results and discussion by referencing particular parts of the results. As suggested, we have changed the title of the “Guiding Principles for PLR” section to “Prospective Consent and Governance Principles for PLR”.

In order to acknowledge former findings on trust/reciprocity in researcher/participant relations, the following citations (mentioned above) have been added:

1. Citation 48: Townsend Anne, Taylor Kim, and Susan M. Cox. (2014). Conceptions of risk regarding a chronic illness survey: Perspectives of participants, researchers, and ethics review board members. IRB: Ethics & Human Research;36 (5):13-20.

2. Citation 11: Cox, Susan M and Michael McDonald. (2013). Ethics is for Human Subjects Too: Participant Perspectives on Responsibility in Health Research. Social Science and Medicine, 98: 224-231.

3. Citation 44: McDonald, Michael, Anne Townsend, Susan M Cox, Darquise Lafrenière, Natasha Damiano Paterson (2008). Trust in Health Research Relationships: Accounts of Human Subjects Journal of Empirical Research on Human Research Ethics, 3 (4): 35–47.

4. Citation 45: Guillemin, Marilys et al. (2018) Do Research Participants Trust Researchers or Their Institution? Journal of Empirical Research on Human Research Ethics. 13 (3): 285-294

5. Citation 41: Kerasidou A. Trust me, I’m a researcher!: The role of trust in biomedical research. Med Health Care Philos. 2017 Mar;20(1):43–50.

6. Citation 42: Guillemin M, Gillam L, Barnard E, Stewart P, Walker H, Rosenthal D. “We’re checking them out”: Indigenous and non-Indigenous research participants’ accounts of deciding to be involved in research. Int J Equity Health. 2016 Jan 16; 15:8.

2.5) Finally, the discussion presents only the case of the US human research regulation. The participants came from 5 other countries in Europe. This is a limitation.

Response 2.5: This is a very good point. We have attempted to make our statements inclusive of both IRB/REB standards. However, as we still focus more heavily on standards in the U.S., we have added the following statement to the end of our limitations statement: “Additionally, this project and writing of this manuscript took place prior to and during the adoption of changing ethical regulations across national borders (i.e., the General Data Protection Regulation or GDPR). We chose to limit our introduction largely to ethical regulatory frameworks in the United States.” A similar statement was added to the bulleted “Strengths and Limitations of this study” list on page 4.

2.6) NB: the references 38 and 39 are inverted.

Response 2.6: This has been corrected, thank you.

VERSION 2 – REVIEW

REVIEWER	Susan Cox University of British Columbia
REVIEW RETURNED	04-Jan-2019

GENERAL COMMENTS	The paper reads very well and makes an important contribution to the literature. I have no further requests for revision.