Reviewer's report

Title: At the cross-roads of participatory research and biomarker discovery in autism: The need for empirical data

Version: 3 Date: 30 April 2015

Reviewer: Sarah Parsons

Reviewer's report:

Thank you for this interesting paper, which deals with an important and timely issue in autism research. I think the paper has merit and the rationale for focusing on CBPR in the context of biomarker research is good. However, I think the paper needs to be strengthened by a more informed understanding of CBPR, including the definition and conceptualisation being applied here. This is important because, as it currently stands, some of the issues raised (particularly around generating knowledge about attitudes to biomarker research) are not equivalent to CBPR. Therefore, claims about whether this scoping review tells us much about CBPR need to be revised. Specifically:

Major compulsory revisions

On p. 4, para 2 the authors state that CBPR is 'one...engagement model' but of course it is not. CBPR really describes a range of approaches through which community participation can be achieved and supported. These approaches can operate in the context of a wide range of research designs, for diverse groups and populations (e.g. see special issue of the Journal of General Internal Medicine: http://onlinelibrary.wiley.com/doi/10.1046/j.1525-1497.2003.30416.x/full). This diversity needs to be reflected more fully in the way that CBPR is described and introduced in the paper and, subsequently defined. The authors provide a definition that seems to draw on a training program rather than published research (WK Kellogg Foundation - this is also not referenced); they then draw upon Israel et al's nine principles to outline what CBPR involves. This is a helpful reference but, of course, not the only way of conceptualising CBPR. Therefore, the authors need to show awareness of the wider field and also justify why they are using these principles in particular in their research.

This matters because on p.8, we are told that the analysis is 'framed within larger CBPR principles' and that the three themes that emerged 'correspond closely with previously articulated principles of participatory research'. This is difficult to judge given that we are not told explicitly which principles are being applied here, and why. Related to this, how was the coding scheme developed? What decisions were made about this and why?

This would also perhaps help to make it clearer why so many articles were excluded from the review. While the inclusion criteria for studies are stated, the
exclusion criteria (and the basis for subsequent exclusion of all but 6 papers from 342 identified) are not. The authors, therefore, need to be more transparent in describing this process of decision-making about the inclusion and exclusion of articles.

Clearer definition and articulation of principles is also needed because simply establishing what a group of respondents ('community members') think about a particular issue is not equivalent to participatory research. That is more of a consultation but it is certainly not participatory, especially not in the sense that the authors themselves suggest relating to the aspiration of more equitable involvement of community members in research. Researchers simply finding out the answers to questions they want to ask does not participatory research make. Consequently, the claims made about the extent to which this paper can really inform us about the feasibility and appropriateness of CBPR to biomarker research need to be revised. The research cited can tell under the first theme can tell us something about parental attitudes, but it cannot tell us about CBPR.

Care also needs to be taken in that first results section (pp.8-9) to describe the results of those studies in a more transparent way. This includes removing any claims that the identification of attitudes was 'systematic' (on what basis is this claim made?). For example, 69% of parents are said to be in favour of genetic testing in the Chen et al study - but this was a qualitative study based on the responses of 42 parents (this needs to be made clear at the very least); the Narcisa et al paper is based on a larger sample but an internet survey - again there are limitations to be acknowledged here. The Trottier paper is based on interviews with 9 participants. Overall, the authors need to present a much more critically aware account of the evidence here, and avoid claims that this evidence amounts to a systematic investigation of parents' views. Related to this, they imply that samples are 'biased' and there is 'misrepresentation of the community' but it is not clear on what basis these conclusions are drawn. Providing a more critical commentary on the research would help to illuminate this.

The same criticality needs to be applied to the second theme (setting of priorities); the Higashijima paper, while interesting, had significant limitations (as acknowledged by the authors themselves) and these need to be acknowledged in this paper too. In addition, the authors should include more coverage of Pellicano et al's research in the UK. It is not clear why this research is not included in this section when it is cited by the authors elsewhere in the paper and does explicitly include relevant discussions relating to research about biomarkers.

Most importantly, Pellicano's research as well as other similar research (e.g. Krahn and Fenton) tends to point community views AWAY from genetics and brain research and TOWARDS service / education based research. This is important because while stakeholders may be supportive of biomarker research if that is all we ask them about, they may not want to prioritise that above other things. So if the authors are really committed to the value of CBPR then how do they envisage reconciling these positions? It would really strengthen the paper if there was some further discussion about the challenges, as well as benefits, of
doing this.

This might even mean that researchers have to think beyond RCTs as the only research design in town - it is possible to do 'good CBPR' using different designs and that is part of the sharing of power that comes with adopting a more participatory approach. By seeing CBPR as a threat to internal validity because there are few RCTs that have been carried out somewhat misses the point; inherent within CBPR is an epistemological challenge about how and where knowledge is constructed. If we are trying to be true to those principles of CPBR then it may mean we need to rethink how research is designed and carried out. The authors' own biases in this respect are made apparent in the middle para on p.5 where they say 'At the extreme, "neurodiversity" proponents even reject the notion that ASD is a condition to be treated...'. This is quite a patronising stance to take - what is the evidence that this view is indeed 'extreme'? And surely, the whole starting point for authentic CPBR is about respecting and valuing different views? This kind of statement is not compatible with such a respectful stance and should be revised accordingly.

Finally, in the Discussion, the authors equate 'funder and public' pressure to political lobbying and censorship of research. These are not the same kinds of things at all and this section needs to be revised. There is certainly a balance to be struck between public views and funding of research but at the moment the bias is very heavily in favour of researchers rather than stakeholders and so there could be a more nuanced awareness shown by the authors about this debate (see Pellicano et al and also Pellicano & Stears on this).

Minor essential revisions

There are typo's and referencing ambiguities that need to be addressed:

p.3. INVOLVE ref (#21) - no traceable source is given for this reference - was it downloaded? If so, from where?

p.4 line 89 - should be beneficiaries

p.4 - where / what is the WK Kellogg Foundation reference? Ideally this needs to be an academic reference rather than a website

p.12 - final paragraph - 'some have argued for...' - who has argued for this?

p. 12, final sentence - hampering the impact of research - what is the evidence for this?

Level of interest: An article whose findings are important to those with closely related research interests

Quality of written English: Acceptable

Statistical review: No, the manuscript does not need to be seen by a statistician.
Declaration of competing interests:

'I declare that I have no competing interests'