Author's response to reviews

Title: Framing overdiagnosis in breast screening: a qualitative study with Australian experts

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Author's response to reviews: see over
Dear Editors,

Thank you for the invitation and opportunity to revise our manuscript entitled “Framing overdiagnosis in breast screening: a qualitative study with Australian experts”.

We would very much like to thank yourself and the reviewers for taking the time to read our manuscript and provide such detailed and useful feedback. It has helped us to strengthen the manuscript. We have made several changes in order to address the reviewers’ concerns. These changes are highlighted in yellow in the manuscript and in our detailed responses to reviewers.

Thank you for considering our revised paper. We look forward to hearing from you.

Kind Regards,

Lisa Parker

<table>
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<tr>
<th>COMMENTS BY REVIEWER #1: Ray Moynihan</th>
<th>Authors’ responses and manuscript changes (Authors’ responses are in italics; manuscript text is in plain text; revised manuscript text is highlighted in yellow)</th>
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<tr>
<td>The research question appears well defined, the methods are appropriate and generally well described, though more detail on how the experts were chosen, and how the interviews were conducted, could be valuable.</td>
<td>We are grateful to both reviewers for highlighting the need for more detail on these points. We have addressed this issue by providing further information on participants and sampling, and on our interview methods. We have also included a Table that shows the characteristics of expert participants.</td>
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Methods

This study is part of a larger Australian National Health and Medical Research Council (NHMRC) funded project examining ethical issues in cancer screening in Australia [20]. One component of the larger project was a qualitative study of contemporary issues in breast cancer screening, using semi-structured interviews with influential breast screening experts. We defined “influential experts” as people working or researching in breast screening who influence the public, primary care practitioners and/or policy makers.
by engaging in one or more of: media commentary; academic or lay publications and presentations; senior service delivery management; membership of government advisory bodies, professional committees and/or non-government/consumer organisations related to breast screening. We sampled purposively from this population, seeking to obtain a wide diversity of views by inviting participants with a range of publicly aired positions [21]. We reasoned that perspectives on screening might be associated with professional backgrounds, so we ensured that we included experts with a range of roles and responsibilities. See Table 1 for further participant details.

We identified potential interviewees by scanning academic and lay literature on breast screening, examining personnel lists on websites of government or non-government advisory and advocacy bodies involved in breast screening, and following up suggestions from colleagues and participants. We used information in the public domain to contact experts by email. Forty-six experts were contacted, and 33 (17 male, 16 female) were interviewed. Thirteen people either did not wish to participate (3), did not respond (9) or were unable to participate in the time available (1). We had a low response rate from senior community advocacy figures. Speculatively, this may have been due to a higher turnover of staff in these (largely volunteer) positions than in other professional roles. That is, the individuals may no longer have been contactable at the email addresses that we had access to. We continued sampling until we had good representation of a range of professional roles and until we reached thematic saturation in our analysis. [22]

LP conducted semi-structured interviews from October 2012 to October 2013, meeting in the participant’s or her own workplace, or talking over telephone if unable to meet in person. The interviews lasted between 39 and 105 minutes (average 66 minutes) and there was no observed difference between face to face and telephone interviews in terms of quality or length [23]. The interviews drew loosely on a set of core questions designed to draw out the participant’s views with regard to breast screening. We also sought to
tailor each interview to the particular expertise and interests of the participants, and explored in depth the leads and topics that arose throughout the discussion [22, 24]. We encouraged the participants to talk about overdiagnosis but did not pre-empt ideas about what was important about this topic. Rather, we began by asking more generally for interviewees’ views. We only pursued particular lines of enquiry about controversial elements – as informed by the literature – if this flowed on from preceding comments of the participant. An additional file outlines sample interview questions (see Additional file 1).

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<tr>
<th>More information on the study limitations are needed</th>
<th>Thank you for pointing this out. We have added further comments on our study limitations in the Discussion:</th>
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<td>More information on the study limitations are needed</td>
<td>As with much qualitative work, we cannot make any predictions about the prevalence or pattern of our results within the wider population, and this may be a useful avenue for future survey research. While this study was limited to the Australian setting, much of the developed world has organised breast screening programs, comparable values, and access to the same body of scientific evidence, and thus the findings are likely to be broadly applicable across these countries. It is possible that experts who participated in our study were somehow different from those who were invited but did not participate. We sought to minimise any bias of this sort by ensuring that we interviewed experts with a range of attitudes to screening, and a wide variety of professional roles and experience.</td>
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I have (below) suggested a re-emphasis in the conclusions.

<table>
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<th>This is addressed in a later section (see below)</th>
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<td>Background: The first paragraph of this section feels too long. It needs to be broken into more than one paragraph, and it needs to be written more clearly – to ensure comprehension by those unfamiliar with the intricacies of this debate. The bulk of the paper is very clearly written – and this Introduction needs to be much clearer</td>
<td>Thank you for these comments. We have re-worked the first part of the Background, clarifying the message and breaking it up into two paragraphs as suggested.</td>
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**Background**

Overdiagnosis in breast screening has become a highly contentious issue and source of strong disagreement amongst experts. In
This paper we use the term “overdiagnosis” to mean the diagnosis through mammographic screening of an asymptomatic breast condition that is non-progressive or so slowly progressive that it would not otherwise have come to the patient’s attention in her lifetime and where this diagnosis provides no net benefit to the patient [1]. The possibility of overdiagnosis in breast screening was acknowledged from its early days of use. The idea that breast screening might lead to the detection of lesions that are “morphologically malignant but clinically benign” was raised as early as the 1970s [2, p490]. Later it was also recognised that mammographic screening would uncover a significant number of in-situ cancers, at least some of which “might not have entered an invasive phase during their lifetime” [3, p14] and would likely fall into the category of overdiagnosis. Despite this, there was limited controversy about overdiagnosis when breast screening programs were introduced in many Western countries during the 1980s and 1990s. This may have been partly because of poor outcomes from treatment of symptomatic breast cancers, and the evidence-based promise of a 30% reduction in population breast cancer mortality.

Since that time, however, the evidence-based estimates of the mortality benefit from breast screening have been revised and reduced [4,5]. In addition, improvements in breast cancer treatment are likely to have further reduced the potential impact of screening in the modern Western setting [5,6]. These developments have fostered a growing interest amongst breast screening experts about the significance of overdiagnosis, which is now a topic of major international concern [7-9].

Page 3- Line 74. It feels like the line “Although it was known that one in five screen detected cancers...” needs a reference. Also, this wording implies...
certainty about the magnitude of overdiagnosis, whereas there is debate about the magnitude, as you outline in this article. I suggest changing wording to reflect the uncertainty, and adding a reference.

| Methods: 1st Paragraph – I feel we need a few more words on how the list of 46 experts was drawn up | We have addressed this comment in the previous response about Methods above |
| Methods: were any steps taken to ensure a range of opinions (eg pro/sceptical) were gathered? | We have addressed this comment in the previous response about Methods above |
| Methods: the supplemental material has only one question on overdiagnosis (forgive me if I have missed something) – which asks - “what are your thoughts on this issue”. If there was any further structure to the questioning on this topic, or some details about the interviewing style/approach, that might be valuable. | Thank you for suggesting that we provide more information on interview questions, prompts, and general style. We have addressed the points on general style in the previous response about Methods above. We have also added greater detail to the supplemental material (Additional file 1) about the interview questions and type of prompts used. |

- (If the topic hasn’t yet surfaced) Recent studies suggest that some cancers found at screening would never have come to clinical attention in that person’s lifetime; for example, Marmot and colleagues suggest that for every 1 life saved by breast screening there are 3 cancers overdiagnosed. What are your thoughts on this issue?
  - Prompt: eg is it – non-existent; existing but not a problem; a problem

- (if appropriate to expert’s views) What level of overdiagnosis do you work with?

- (if considered a problem) Should screening programs take any responsibility for reducing overdiagnosis? E.g. should we tailor the program to minimize overdiagnosis?

- (if expert talks about the development of biomarkers or other prognostic tools as a way of addressing concerns)
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<tr>
<th><strong>Discussion:</strong> Apart from an indirect reference to this being an Australian study, there is no discussion of Limitations – please include some text on the limitations as you see them.</th>
<th><strong>What should we do in the meantime?</strong></th>
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<td><strong>We have addressed this comment in a previous response (see above)</strong></td>
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| **Conclusion:** Page 18 – Lines 433-436. I would be less inclined to offer this set of recommendations for how people conduct themselves in public debates, and more inclined to summarise the results of your important study. If you feel the recommendations (or some of them) are justified, I feel you need to make the case more clearly in the Discussion for why your results demand these sorts of recommendations. For example, I feel that suggesting that people “who engage in future debates”, nominate whether “their frame is commensurable or incommensurable with others” is not very helpful, in that opportunities to engage in debate are often very limited (in time or space). My sense is that you have uncovered some very useful material on how different people frame this problem (and potential solutions), and that the recommendations for how folks conduct themselves in debates are in some ways a little disconnected from these findings. | **This was a useful comment and we have re-worked the conclusion to address the reviewer’s concerns.** |

**Conclusions**

Our results demonstrate that experts approach overdiagnosis in various ways, see a range of issues and values at stake, and are inclined to promote different solutions. This may be an important contributor to the ongoing controversy in this topic, and offers a new explanation for why some debates about overdiagnosis are so heated. The concept of experts using different frames when thinking and talking about overdiagnosis might be a useful tool for those who are engaged in the topic, assisting with communication and facilitating better understanding of others’ viewpoints.

| **Page 16, Line 396 – I would suggest removing the word “greatly”** | **Yes we have removed the word “greatly”** |

**COMMENTS BY REVIEWER #2: Paolo Giorgi Rossi**

**Objective:** the two questions do not help to understand what the authors are going to answer. Question 1) discuss with whom?  

The second question seems less linked to framing and more with the knowledge about over diagnosis, except for value

**Thank you, we have re-worded the first research question to provide more clarity.**  
- **How do Australian breast screening experts frame overdiagnosis?**

**Thank you for this comment. Frame analysis is a common analytic method in social science research. The dimensions of framing**
judgements.

that we refer to in this research question were informed by our reading of the framing literature, and are used in standard framing analysis (see references by Entman [1993] and Ryan [1991] in our reference list). We believe that we have justified this approach sufficiently and linked it to the most useful relevant literature, but are happy to take further instruction from the editor on this point.

**Methods:** Please describe the interview conduction: how was conducted the interview?

Thank you for this comment about Methods; please see our response to Reviewer 1 on the same topic.

**Methods:** Which questions? If there were no fixed questions please explain how the interviewers agreed to standardise the interview.

Thank you for this comment about Methods; please see our response to Reviewer 1 on the same topic.

Please also note that this was a semi-structured, not a structured interview. In semi-structured qualitative interviewing, questioning is responsive to the particular participant rather than being standardised across all interviews. There is a guiding structure, but the interviewer follows up on leads, or asks additional questions to better understand the individual’s point of view. The focus is on achieving the best possible information from and understanding of the participant, rather than asking exactly the same questions of each participant.

**Results:** The paragraph “how experts used frames” should give information in a more systematic way. May be a table with all the experts and the frames they use (may be in a qualitative way such as “+” “+/-” and “-“) and the expert’s role. I think this is very interesting.

This is an interesting suggestion. We examined the data closely for any patterns in how experts used overdiagnosis frames, and touched on this briefly under “How experts used frames” in the Results. Prompted by this comment, we have added an extra paragraph to this section and added Additional File 2 to provide tables showing the two patterns identified (relationship between expert’s professional roles and the overdiagnosis frames used; and relationship between overall views about breast screening and the overdiagnosis frames used). The new paragraph reads:

There were observable patterns between experts’ overall views on breast screening and their use of overdiagnosis frames. All experts who were
Critical of breast screening used the “don’t hide the problem” frame, and none of them used the “stop squabbling in public” frame. Experts who were supportive of breast screening used one or other, but not both, of these frames (in approximately equal numbers), and were the only group to use the “stop squabbling in public” frame. Further detail on this is available in Additional file 2).

Can you give an idea of any gender difference in the use of frames?  

We did not find any obvious gender difference – except that all the consumer advocates were women, and all the consumer advocates used frame 2. Our analysis suggested that the use of frame 2 was more likely tied to professional role than gender, and we did not pursue the issue of gender in the text.

Discussion: In the discussion there is no mention of what are the strengths and limitations of this approach and of this study in particular. I mean how the results corroborate the method or not, which results are surprising or may give an idea of how much the controversy is bipolarised or is also interiorised by many experts.

We have provided further detail in the strengths and limitations section of the Discussion as per our previous response to Reviewer 1.

We weren’t entirely clear about the second part of this comment but we hope the revised text address these concerns. We are happy to be guided by the editor here.

Discussion: I agree not to discuss how these results should suggest practice or other practical implications because this is just a tool to analyse the question and each actor should use it in a different way.

Thank you for this comment about the Conclusion; please see our response to Reviewer 1 on the same topic

Results: Page 10: please explain with some notes what does it mean to be from East Sydney or North Sydney.

Was this point raised about stopping active invitations? Because active invitation has been shown to be effective in reducing inequalities.

This was a very helpful comment. We have re-worked the quotation to make the meaning clearer for all readers:

“There’s probably people in [suburbs of lower socioeconomic status] who stop going to screening. Because they’re not as sophisticated … and they come from non-English speaking backgrounds. The message they get is that screening is not needed… It’s okay if you’re in the [suburbs of higher socioeconomic status] because you’ll keep coming anyway.” (Expert #29, clinician)

This quote referred to experts’ concerns that
negative publicity might deter women from attending screening (as noted in the text immediately prior to the quote). There was no discussion about stopping invitations to screening.