

PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	Outcomes valued by people living with dementia and their care partners: Protocol for a qualitative systematic review and synthesis
AUTHORS	Booi, Laura; Wheatley, Alison; Brunskill, Greta; Banerjee, Sube; Manthorpe, Jill; Robinson, Louise; Bamford, Claire

VERSION 1 – REVIEW

REVIEWER	Jenny van der Steen Leiden University Medical Center, Public Health and Primary Care
REVIEW RETURNED	06-Apr-2021

GENERAL COMMENTS	<p>The proposed review will synthesize perspectives of persons with dementia and family caregivers on important outcomes from their perspectives and fits with contemporary views on person-centered care and soliciting views and participation of persons with dementia themselves. A qualitative synthesis makes sense and the manuscript is well written. I have a questions about the added value compared to recent reviews, and some minor points that may be clarified.</p> <p>Major point What is the added value compared to recent reviews exactly? I'm referring to the paragraph that starts with "Qualitative studies of outcomes valued people living with dementia have been synthesised in two recent reviews.(13, 14) However, our initial searches identified several relevant papers that were not included." Morrisby et al. (13) and Tochel et al. (14) included qualitative studies and also studies with other designs, while the proposed study will include qualitative studies only to arrive at a qualitative synthesis. Perhaps the two recent reviews include a broader range of studies but the searches were less sensitive with respect to studies with a qualitative design. However, it is not clear why a qualitative synthesis would improve by excluding studies with other designs because various study designs can be covered in a qualitative synthesis such as often done in scoping reviews. Also, the statement about the other reviews having missed records is not convincing without reference to the respective search strategies and how these would differ not only in sensitivity but also in specificity. The next sentences in this paragraph, on the other hand, provide a good rationale for another review on essentially the same topic: "Further, we have not found parallel syntheses of the views of care partners on outcomes desired for themselves or for the person with dementia they support. If possible, we will also include papers</p>
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	<p>reporting on the outcomes that people with dementia identify as important for their care partner.”</p> <p>It would be even more convincing that there is important information that has been overlooked if the authors mentioned some examples of studies included in the other reviews while the relevant data on the exact perspectives has been ignored by any other work so far. You may wish to look at McCauley R et al. who just published in Palliative Medicine a review on mutual support between patients and family caregivers, and there was no study about dementia which suggests little is known about how persons with dementia feel about supporting their family caregivers, in e.g., alleviating distress or taking away from caregiver burden. I would expect this to be a nice niche for your review.</p> <p>Finally, in your next sentence, the additional value should be clarified:</p> <p>“The aim of this qualitative synthesis is to add to the existing literature by systematically searching for papers exploring the related concepts of outcomes, well-being and quality of life.”</p> <p>That is, Tochel et al. (ref 14) used several search terms for quality of life too. In all, given the extensive recent reviews, the rationale for another one should be crystal clear and therefore needs a stronger justification.</p> <p>Minor points</p> <p>Title and abstract could be clarified in regards what outcomes are being addressed (not necessarily outcomes for research, probably more so outcomes to evaluate policy and services)</p> <p>The introduction starts off with UK and international, but the references cited are exclusively UK and the first reference is incomplete (Department of Health in which country). You may wish to avoid an impression of UK (assumed readership) bias.</p> <p>Aim: the broad range of studies is inconsistent with the limited inclusion of qualitative studies only.</p> <p>In the methods, please provide more detail on how you would translate lived experiences into outcomes in the analysis of the data.</p> <p>The assessment of quality developed by Croucher et al. (2003) needs more explanation as it is not well known and the reference is about its application in research on homeowners. Please also explain why this is the “most appropriate” among the approaches referenced.</p> <p>Data synthesis: how would you compare the outcomes identified as important by the different partners in a rigorous manner, for example, would you develop codes for the two perspectives independently first?</p> <p>How would you avoid assessing coherence as evaluating the quality of your own synthesis?</p> <p>In the discussion, you may address the exclusion of quantitative measures as a limitation as mixed methods often provide rich data</p>
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	about what is important and to how many persons this is important which is, in the end, relevant as it comes to measuring outcomes that matter.
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REVIEWER	Rachel Herron Brandon University, Department of Geography
REVIEW RETURNED	11-Apr-2021

GENERAL COMMENTS	<p>The protocol paper reports on a study that will synthesize outcomes identified as important from the perspectives of people with dementia and their care partners. The authors provide a succinct and persuasive overview to justify their study and the need to understand outcomes from the perspective of people living with dementia and partners in care. I was particularly impressed with the involvement of the mixed stakeholder group in the research design, interpretation, and decisions about dissemination. I thought maybe this information should come earlier in the protocol since this group was instrumental in the protocol design. The stakeholder group will contribute to the relevance and appropriate application of the research findings, pushing the field and knowledge mobilization forward. I have some other minor revisions and clarifications for the authors to consider before the article is accepted.</p> <p>The third strength in the strength and limitations section needs rewriting for clarity</p> <p>On the first page the word “by” is missing from the sentence “Qualitative studies of outcomes valued people living with dementia have been synthesised in two recent reviews.(13, 14)”</p> <p>It would be helpful if the researchers would elaborate on the statement “Separate search terms have not been included relating to care partners, since relevant papers will be captured within the broader search.” What are some examples?</p> <p>It seems like the researcher will search for well-being, quality of life, and lived experience. A lot of relevant research may not use the words well-being or quality of life. What other search terms will be used to help ensure all relevant literature is found?</p> <p>On page 10, there is a word missing from the sentence “Following detailed review of published approaches (30, 31, 33-36) we identified the developed by Croucher (2003) as most appropriate to our review.(37)”</p>
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VERSION 1 – AUTHOR RESPONSE

Reviewer 1’s major points	
<p>What is the added value compared to recent reviews exactly? I’m referring to the paragraph that starts with “Qualitative studies of outcomes valued people living with dementia have been synthesised in two recent reviews.(13, 14) However, our initial searches identified several relevant papers that were not included.”</p> <p>Morrisby et al. (13) and Tochel et al. (14) included qualitative studies and also studies with other designs, while the proposed study will include qualitative studies only to arrive at a qualitative synthesis. Perhaps the two recent</p>	<p>Thank you for this feedback. We have revised the introduction in light of your comments, describing the added value of this review compared to recent reviews, and strengthening our argument for why this review is needed.</p> <p>Our rationale for focusing on qualitative studies has also been elaborated. Quantitative studies provide data on priorities given to pre-specified checklists or measures; the extent to which these capture the full range of outcomes valued by people living with dementia and their</p>

<p>reviews include a broader range of studies but the searches were less sensitive with respect to studies with a qualitative design. However, it is not clear why a qualitative synthesis would improve by excluding studies with other designs because various study designs can be covered in a qualitative synthesis such as often done in scoping reviews.</p>	<p>care partners is not known. By focusing on studies using qualitative methods, which allow people living with dementia and their care partners to articulate desired outcomes in their own words without any constraints, we can be confident that the findings will capture the full range of outcomes valued. Articulating outcomes can be challenging, particularly if there is a focus on outcomes of specific services, where people living with dementia and care partners may have low expectations or be unaware of the wider range of outcomes that could be achieved from a comprehensive package of support. For this reason, we will also include papers on lived experience, since we believe these will shed additional light on valued aspects of life which may otherwise be missed.</p>
<p>Also, the statement about the other reviews having missed records is not convincing without reference to the respective search strategies and how these would differ not only in sensitivity but also in specificity.</p>	<p>Key differences compared with the earlier reviews by Tochel (1) and Morrisby (2) are that we are not restricting papers to:</p> <ul style="list-style-type: none"> - specific dementia subtypes (as in Tochel) - those explicitly including care partners (as in Morrisby) - those living in the community (Morrisby) - those relating to service utilisation (Morrisby) <p>This is now described further in the methods section.</p>
<p>The next sentences in this paragraph, on the other hand, provide a good rationale for another review on essentially the same topic: "Further, we have not found parallel syntheses of the views of care partners on outcomes desired for themselves or for the person with dementia they support. If possible, we will also include papers reporting on the outcomes that people with dementia identify as important for their care partner." It would be even more convincing that there is important information that has been overlooked if the authors mentioned some examples of studies included in the other reviews while the relevant data on the exact perspectives has been ignored by any other work so far.</p>	<p>Previous reviews have not clearly distinguished between outcomes for people living with dementia and care partners. This is illustrated in quotations in the published reviews which could be interpreted as outcomes for people living with dementia and/or care partners. For example, the following quote from a spouse has been interpreted as an outcome for the person living with MCI ('maintenance of patients' identity and personality'):</p> <p><i>"My best friend [husband with MCI] has gone. He is part of me, but he is no longer the same person. I really miss him"</i>. (Tochel, 2019, pg 243).</p> <p>We do not know the extent to which the person living with MCI would share this sentiment, therefore interpreting it as an outcome for him is questionable. We would interpret this as an outcome valued by the care partner herself, since there is a clear sense of missing the connection with her husband. We feel this illustrates the importance of clearly separating outcomes valued by people living with dementia and care partners.</p>
<p>You may wish to look at McCauley R et al. who just published in Palliative Medicine a review on</p>	<p>Thank you for this suggestion; we have incorporated this paper into our argument for the</p>

<p>mutual support between patients and family caregivers, and there was no study about dementia which suggests little is known about how persons with dementia feel about supporting their family caregivers, in e.g., alleviating distress or taking away from caregiver burden. I would expect this to be a nice niche for your review.</p>	<p>purpose of the review.</p>
<p>Finally, in your next sentence, the additional value should be clarified: "The aim of this qualitative synthesis is to add to the existing literature by systematically searching for papers exploring the related concepts of outcomes, well-being and quality of life." That is, Tochel et al. (ref 14) used several search terms for quality of life too. In all, given the extensive recent reviews, the rationale for another one should be crystal clear and therefore needs a stronger justification.</p>	<p>While Tochel et al (1) did use a range of search terms relating to quality of life, we have introduced a number of additional synonyms for outcomes, quality of life and wellbeing in addition to lived experience papers (as is now shown in table 1). We have not included the more technical terms (e.g. HRQOL) used by Tochel as these are unlikely to be used in qualitative papers.</p> <p>Moreover, Tochel et al (1) focused their search on Alzheimer's Disease and/or MCI, meaning that data about other dementia subtypes may not have been captured. We have attempted to make our dementia search as broad as possible in order to incorporate perspectives of people with rarer dementias and their care partners.</p> <p>Similarly, Morrisby et al's (2) search terms were also more restrictive than our intended terms; for example, they excluded PLWD in care homes.</p>
<p>Review 1 minor points</p>	
<p>4. Title and abstract could be clarified in regards what outcomes are being addressed (not necessarily outcomes for research, probably more so outcomes to evaluate policy and services)</p>	<p>Thank you for this suggestion. We intend our review to cover 'outcomes' in the broadest sense, rather than focusing on what is provided by services. We have clarified the definition of outcomes in the introductory section and in the abstract, but have not made any changes to the title</p>
<p>5. The introduction starts off with UK and international, but the references cited are exclusively UK and the first reference is incomplete (Department of Health in which country). You may wish to avoid an impression of UK (assumed readership) bias.</p>	<p>Thank you for this. We have now incorporated more non-UK literature, including national dementia strategies, OECD policy documents, and the Global Dementia Charter from Alzheimer's Disease International</p> <p>The incomplete reference has also been updated.</p>
<p>6. Aim: the broad range of studies is inconsistent with the limited inclusion of qualitative studies only.</p>	<p>The aim of the review is to synthesise outcomes articulated by people living with dementia and their care partners in their own words, rather than using pre-populated instruments. Qualitative methods are therefore most appropriate for capturing this kind of data. We are nevertheless including a range of qualitative methods where appropriate, including</p>

	observation.
7. In the methods, please provide more detail on how you would translate lived experiences into outcomes in the analysis of the data.	<p>Thank you for raising this issue. The extent to which outcomes are explicitly articulated varies between papers, particularly those relating to lived experiences. Our intention is to use the thematic approach described by Braun and Clarke (3) and to start by reading and rereading a sample of papers (which will be selected to ensure the inclusion of some lived experience papers) and generate initial ideas about themes and outcomes from the data. This will involve interpreting lived experience data through an outcomes 'lens', for example looking for terminology that can be interpreted as expressing a desired outcome (e.g. 'want', 'need', 'wish') or identifying an element of post-diagnostic support that is missing; at this stage, our aim will be to stick closely to the terms used in papers and avoid imposing concepts on the data. Initial codes will be discussed in data workshops to produce a preliminary list of potential outcomes.</p> <p>This process will continue iteratively until the dataset has been analysed. As the analysis proceeds through further discussion and the development of memos delineating and summarising specific codes, we anticipate that more abstract, conceptual codes will be generated as we explore the relationship between our original themes.</p> <p>We have added this information to the text.</p>
8. The assessment of quality developed by Croucher et al. (2003) needs more explanation as it is not well known and the reference is about its application in research on homeowners. Please also explain why this is the "most appropriate" among the approaches referenced.	<p>We have amended the text to explain this.</p> <p>We are following the approach to qualitative synthesis used by Thomas and Harden (4), in which quality appraisal criteria are adapted to the subject area. We examined several methods for quality assessment, mapping headings across tools, and tested them on a small batch of papers. From this exercise, we identified some elements that were less relevant to our review (e.g. they were not reported in the papers of interest) and others that were difficult to operationalize. Based on this, we chose Croucher as being the easiest to operationalize, covering the key quality issues of relevant to our review and containing few superfluous items.</p>
9. Data synthesis: how would you compare the outcomes identified as important by the different partners in a rigorous manner, for example, would you develop codes for the two	<p>The intention is to develop a single coding framework. However, as part of our analysis we will use NVivo functionality to code: a) who the outcome is for and b) whose perspective it is</p>

<p>perspectives independently first?</p>	<p>from. This will allow us to examine the similarities and differences in emphasis between and among people living with dementia and their care partners.</p>
<p>10. How would you avoid assessing coherence as evaluating the quality of your own synthesis?</p>	<p>We will assess coherence using the approach described in Colvin et al (5) in the series of papers outlining the CERQUAL approach. Since the entire findings sections of included papers will form the data for the synthesis, this will ensure that all data relevant to the review finding are available for scrutiny. As themes (or outcomes) emerge, we will examine papers to identify whether these are consistent across all included papers and document contradictory findings. We will specifically examine whether themes are consistent across papers focusing on lived experience vs those with a more explicit focus on outcomes. Further we will explore the extent to which themes identified fit with papers on seldom heard populations to ensure that additional themes that may be specific or less relevant to certain populations (e.g. those with young onset dementia; spousal care partners; people from minority ethnic groups) are captured. As part of our qualitative approach we will also look for plausible alternative ways of grouping the themes; the coding and analysis will undergo a number of iterations to enable us to find the most coherent and parsimonious way of summarising outcomes. Reflexivity is (or should be) part of any qualitative analysis. One advantage of the CERQUAL approach is that the coherence of <i>each</i> review finding will be evaluated. Thus rather than providing a global statement about the coherence of the findings, the CERQUAL approach will help us be explicit about how and why judgements about the coherence of individual review findings have been made.</p>
<p>11. In the discussion, you may address the exclusion of quantitative measures as a limitation as mixed methods often provide rich data about what is important and to how many persons this is important which is, in the end, relevant as it comes to measuring outcomes that matter.</p>	<p>Our focus is on outcomes valued by people with dementia and their care partners, expressed in their own words. The inclusion of quantitative studies would potentially skew the results, since they report information on prespecified outcomes. Since these may not be the same as those defined by PLWD and care partners, we think it essential to exclude these papers. However, mixed methods studies will be included if they contain a qualitative component that meets the inclusion criteria.</p>

Review 2 minor points	
1. The protocol paper reports on a study that will synthesize outcomes identified as important from the perspectives of people with dementia and their care partners. The authors provide a succinct and persuasive overview to justify their study and the need to understand outcomes from the perspective of people living with dementia and partners in care.	Thank you.
2. I was particularly impressed with the involvement of the mixed stakeholder group in the research design, interpretation, and decisions about dissemination. I thought maybe this information should come earlier in the protocol since this group was instrumental in the protocol design. The stakeholder group will contribute to the relevance and appropriate application of the research findings, pushing the field and knowledge mobilization forward. I have some other minor revisions and clarifications for the authors to consider before the article is accepted.	Thank you for the suggestion. On balance, we have decided to leave the PPI section where it is.
3. The third strength in the strength and limitations section needs rewriting for clarity	Thank you. Following comments by the other reviewer and editor, the strengths and limitations have been re-written and this one is no longer included.
4. On the first page the word "by" is missing from the sentence "Qualitative studies of outcomes valued people living with dementia have been synthesised in two recent reviews.(13, 14)"	Thank you. This has been corrected.
5. It would be helpful if the researchers would elaborate on the statement "Separate search terms have not been included relating to care partners, since relevant papers will be captured within the broader search." What are some examples?	We have elaborated this in the text and hope it is now clear. Essentially, papers relating specifically to care partners of people with dementia will be a subset of all papers retrieved by the existing search (for example, a paper with the title 'carers for relatives with dementia' would be picked up by a search for 'dementia'; the additional term 'carers' is not needed). Therefore, adding a specific term would not identify any additional papers, but would potentially exclude other relevant papers.
6. It seems like the researcher will search for well-being, quality of life, and lived experience. A lot of relevant research may not use the words well-being or quality of life. What other search terms will be used to help ensure all relevant literature is found?	The full list of terms is given in Appendix 1 in Medline format and we have added a table to show these in a more readable format (table 1). The terms were developed through examining both published search strategies and known papers of interest to ensure they would be captured, as well as identifying additional synonyms for the terms used. We believe that the list is now broad and comprehensive, covering a range of related words for both outcomes and

	<p>lived experience.</p> <p>However, since we are focusing on qualitative papers exploring the perspectives of PLWD and care partners, we have not included more quantitative terms (e.g. HRQOL) which have been used in some previous reviews.</p>
<p>7. On page 10, there is a word missing from the sentence “Following detailed review of published approaches (30, 31, 33-36) we identified the developed by Croucher (2003) as most appropriate to our review.(37)”</p>	<p>Thank you. This has been corrected.</p>

VERSION 2 – REVIEW

REVIEWER	Jenny van der Steen Leiden University Medical Center, Public Health and Primary Care
REVIEW RETURNED	29-Jun-2021
GENERAL COMMENTS	Upon suggestions and questions of the reviewers, the authors have improved scientific rigor and clarified justification of the review. I look forward to see relevant results.