Author's response to reviews

Title: Extending life for people with a terminal illness: a moral right and an expensive death? Exploring societal perspectives

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Version: 3 Date: 23 December 2014

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

RE: Extending life for people with a terminal illness: a moral right and an expensive death? Exploring societal perspectives

Thank you for the comments relating to our submitted manuscript. We, the authorship team, are delighted at the positive response and comments that the paper received.

Responses to both reviewers’ comments are detailed below. Our responses follow the corresponding comment from the reviewers’ report. When a specific line number is referred to this relates to the clean copy of the revised manuscript i.e. without track changes.

In addition to those revisions and clarifications requested by reviewers, we have made some additional minor textual edits that we noted up as we revised the text. These amendments have been made where they improve clarity and do not represent any significant re-writing. Furthermore, the factor labels have been changed. This was not a request from our reviewers but follows comments made at a recent research engagement event in which it was felt they could be better phrased.

We look forward to hearing your response.

Yours sincerely,

Neil McHugh
Reviewer's report

Title: Extending life for people with a terminal illness: a moral right or an expensive death? Exploring societal perspectives

Version: 2 Date: 18 October 2014

Reviewer: Amanda Wolf

Reviewer's report:

Minor Essential Revisions

1. Provide an abstract

An abstract had been uploaded at the time of submission although it was not contained within the main body of the paper. This has now been included in the main word document – page 2.

2. Include a statement about ethical approval/s.

A statement has been added lines 186-188.

3. In the Methods section, detail is provided on the statement set and the participants. There is a need to clarify that the statement set was selected with attention to ensuring a representative sample of the ‘population’ of opinions relevant to the study. Researchers should be convinced that the statement set is as fully ‘saturated’ as far as they can judge, and that there are no known relevant opinions that are not contained, more or less, in the set. However, Q methodology’s principles for selecting participants (from whom to collect Q-sort data) need to be more carefully described, without attributing the ideal of representativeness and saturation to the selection of sorters (although the methods actually used were entirely appropriate). Q methodology does not proceed by iterative data collection and analysis, ceasing only when no new views are discovered.

The reviewer makes good points about our explanation regarding derivation of the Q set (statement set) and respondent sample. Whilst we carefully and rigorously searched for statements for our Q set from several sources, identifying a range of key informants, and so can be relatively confident we did not exclude something important, one can never be absolutely certain that further searching would not unearth a new statement. We have revised the methods section to address the points raised and think that the principles of sampling both statements and participants are now clearer in the text. Please see revised methods section (in particular ‘The statement set’ and ‘Data collection sections’).

4. In the first paragraph of the Results, the inclusion of the two general public factors is explained. It is misleading to state that this ‘accounts’ for the views of the general population. Rather, the
‘meta’ Q sorts take their place vis-à-vis the views of the purposive sample, and serve mainly to position them.

We agree this was a misleading term and have revised accordingly (see first paragraph of results section).

5. Tables 2 and 3 make clear that ‘exemplars’ are defined by the flagging method, giving the 19, 15, and 16 exemplars noted in paragraph 2 of the results section. However, based on the text in that paragraph, there should be fewer exemplars, as some of the flagged sorts do not meet the condition of ‘highly positively associated with one factor and negatively, or hardly, associated with the other two accounts’ – that is, they are ‘mixed loaders’. The statistical measure for ‘highly associated’ is correctly reported (0.37); thus EX005 and 14 others amongst the three factors should not be counted as ‘exemplars’.

We agree with the reviewer’s comment that the explanations for ‘exemplars’ and ‘mixed loaders’ are not clear here. Correspondingly we have altered the text in paragraph 2 of the results section to highlight that the flagging method is used to indicate defining sorts and that this method relates to the definition of a ‘mixed loader’ (see lines 314-319 and Table 2). Furthermore, to avoid confusion we now use the term ‘defining sort’ in place of ‘exemplars’ throughout the paper and in Tables 2 and 3. These changes to not affect the number of defining Q sorts (19, 15, and 16) we find. For further clarity significant loadings in Table 2 are now shown in bold type.

6. In the Results, paragraph 3, two statements are noted as ‘consensus’ statements; however in the last paragraph of the Factor 1 description, a third statement (#8) is also described as a consensus statement. Clarify the use of ‘consensus’ in these two places.

The use of consensus in the Results, paragraph 3 refers to statements that do not distinguish between any two pairs of factors (non-significant at p>0.01), as noted at the bottom of Table 1. This is the sense in which the term is typically used in Q methodology.

In the last paragraph of the Factor 1 description (to describe statement #8) we meant to highlight that this statement is placed in a similar position across the three factors. Although it does not satisfy the statistical test for consensus (it is in fact distinguishing for F3) – it does seem worth noting that the factor scores are 4, 5, and 5*for F1, F2 and F3 respectively All factors agree strongly with statement #8, although some more strongly than others. We accept that our use of the term consensus here is misleading as it does not refer to the same definition as outlined in the previous paragraph. As such we have changed this term to ‘agreement’ to clarify this issue (line 379).

7. In the second paragraph of the Factor 2 description, a claim is made that ‘extending treatments is considered wrong’. But statement 29 does not (on its own) suggest anything more than a definitional matter (that not giving access to medicines is the same as killing).

We agree with the reviewer that a claim that ‘extending treatments is considered wrong’ cannot be drawn from the positioning of statement 29 alone – Not giving access to life-extending medicine to a
person with a terminal illness is the same as killing them. It was our belief that this claim can be made by considering the account of Factor 2 as a whole and this was our intention in making this claim. On reflection, however, we see F2 as concerned more with the moral right of providing treatment (#27, +3*) than the moral wrong of withholding treatment (#28, +1*) (although they are clearly related issues). We have amended the factor description to omit the claim related to statement #29 and to add some clarity to our interpretation of factor 2 (see second paragraph of Factor 2).

8. The Ethical and Policy Implications are based on arguments about the way ‘public opinion’ can inform decisions. Since the study has used non-representatives (experts), some justification is needed for taking their views as indicative of the public (especially as Factor 3 did not have a parallel in the general public study).

This is an interesting issue which is now more clearly addressed in the discussion in an additional section immediately following the sub-header “Ethical and policy implications”. Whilst we accept that others might take a different position, ours is that the nature of societal perspectives is best addressed by purposive sampling methods and in-depth data generation techniques. Once identified, a number of steps are needed before policies can be written which reflect on public values. Support for those views identified in the wider population can be measured using survey techniques; deliberative methods can be used to debate the quality or application of perspectives in public policy; and/or normative analysis might be brought in. We hope this position is now more clearly expressed.

Discretionary Revisions

1. The title phrase ‘a moral right or an expensive death’ can be omitted; it could lead a reader to expect something more comparative than the study that is reported.

Since this revision is discretionary, we have altered only slightly, adding ‘and’ instead of ‘or’.

2. There is some debate as to how to classify Q methodology, whether as ‘mixed methods’ or as a unique approach that uses both quantitative and qualitative data. Both claims are made (last paragraph on the first section in Background and first paragraph of methods). I suggest omitting mention of mixed methods.

Reference to Mixed methods has been deleted (line 128).

3. The Background section presents an argument that NICE’s perception of public support for special life-prolonging treatments may not have been a fully accurate reflection of the views of its Citizens Council (and the public). This same section then notes an analysis of the support offered by some in the supplemental guidance consultation that seemed to rely on the lack of fully capturing the value of end-of-life extensions (not unlike aspects of the interpretation of Factor 3). Later, the authors suggest that NICE should ‘provide a transparent account of how public values are incorporated into processes’ (Ethical and Policy Implications, paragraph 2). While I find the
need for transparency persuasive, the authors could clarify (a) that there is reason to question NICE’s (earlier) interpretation of societal values (and that this may simply be due to a lack of good evidence for them, hence the value of the study that is reported); (b) that over and above transparency, there may be a need for better understanding of what accounts for support/lack of support for NICE’s guidance (again a contribution of the study).

NICE social values means the SVJ document published by NICE which is partly based on the views of the public as represented through their Citizens Council. There is a need, as stated by the reviewer, for more information about the nature of societal values. We have added a line to the background section stating this case and linking it to the objectives of our study (see last paragraph of background section).

4. In Data Collection, it would be helpful to see the range of academic disciplines in the set of participants.

We agree with the reviewer’s comment. This information is already included within Table 2 but we have now added a sentence to the end of the first paragraph in the Data Collection section to alert readers to this fact. We had to swap the positions of Tables 2 and 3 to correspond to when they are referred to in the text (all subsequent mentions of table 2 and 3 have been updated).

5. The ‘purposive sample’ may be better named ‘expert sample’ in part to describe the participants more accurately and in part to contrast with ‘general sample’

We debated this term over time and in a previous version of this paper the ‘purposive sample’ was called the ‘expert sample’. However, while participants have been included because of their expertise in the area of end of life through their professional life other participants have been included because of experience gained in a personal capacity. The latter group could still be described as expert, but we felt on balance that the label tends to imply professional expertise and settled on ‘purposive’ to describe individuals selected because of their rich perspectives.

6. In a few places, factor interpretations are described as narratives (for example in the first sentence of the Factor 3 discussion). It might be helpful to clarify in the introduction to the factor presentations, in what sense they can be considered as narratives.

In describing factor interpretations as ‘narratives’ our intention was only to use this word as an interchangeable alternative to ‘accounts’ – and an attempt to capture the ‘wholeness’ of Q factors, their interpretation and their representation as descriptive pieces of writing – the telling of a story. However to avoid confusion with other definitions and uses we have substituted the word narrative for account throughout the paper, so making consistent our use of terms.
Minor issues not for publication.

1. Minus sign

We are unsure about this as we thought we had already used a typographical minus sign instead of a hyphen.

2. Numerals

We agree with the reviewer and have made the format consistent to numerals throughout.

3. Email typo

The typo in Cam Donaldson’s email has been fixed (line 35).

4. In-text references

References have been changed to The BMC Medical Ethics reference style.

5. Use of HTA

We agree with the reviewer and have removed the abbreviation referring to HTA throughout the text and in Table 2.

6. SVJ

We agree with the reviewer and made ‘social’ lower case (line 133) and removed the abbreviation of SVJ throughout.

7. Grammatical problem

We have fixed the grammatical problem (line 170).

8. Each . . . . is

This grammatical error has been changed as per the reviewer’s comment.

9. Different . . . . from

This grammatical error has been changed as per the reviewer’s comment (line 475).

10. Close other

Amended (line 507).
Level of interest: An article whose findings are important to those with closely related research interests

Quality of written English: Acceptable

Statistical review: Yes, and I have assessed the statistics in my report.

Declaration of competing interests: I declare that I have no competing interests
Reviewer’s report 2

Title: Extending life for people with a terminal illness: a moral right or an expensive death? Exploring societal perspectives

Version: 2 Date: 11 November 2014

Reviewer: Kristin Bakke Bakke Lysdahl

Reviewer’s report:

Reviewer’s report on ‘Extending life for people with a terminal illness: a moral right or an expensive death? Exploring societal perspectives This is a study addressing the important issue of resource allocation in health care. A comprehensive mixed methods approach is use to investigate UK societal perspective in relation to the relative value of life extension for people with terminal illness, which should be of interest beyond the UK setting. Overall, the paper is thoroughly prepared and only a few critical comments are provided below.

Minor Essential Revisions

1. The method used in the study is generally well described. However, some more details about how the statement sets was design could have been provided (in the methods section or in an appendix), e.g. regarding the how the interviews was performed, background characteristics of the persons interviewed etc.

Further detail has now been added. See revisions to methods section on pages 5, 6 and 7. Table 2 and 3 summarise respondent characteristics.

2. The factors are stated to represent three distinct perspectives, which may seem confusing when the correlation between factor 1 and 2 is 0.68 (Results, third paragraph).

This is a relevant point and although we refer to the issue of similarity in the paper, the word “distinct” has now been deleted. Although we regard factor 3 as sufficiently distinct to warrant inclusion, it has some features in common with factor 1. For example, Factor 3 supports value for money – which is also a central issue for factor 1 but for factor 3 value is regarded more broadly than simply gains in health and is partly defined by individual (over societal) preferences. Additionally factor 1 believes there should be no special cases whereas in factor 3 there are cases when expensive life-extending treatments should be provided (contingent on quality of life being good). That said we agree that ‘distinct’ is not the most appropriate word to use.

3. Limitations of the work are stated, regarding generalization, and most importantly regarding the policy implications. Further discussions of the methods used would improve the article, e.g. regarding the coverage of the statement sets, the added value of the audio-recorded interviews after completion of the Q sort. Did the latter e.g. contribute to understand reasoning behind the importance of both individual choice and preciousness of life in factor 2?
We have added detail about the derivation of the statement set (pages 5 and 6) and use of audio recordings (page 7). The audio recorded interviews did indeed help in our interpretations and quotes are taken verbatim from these interviews and included in our factor descriptions. Preciousness of life and choice are findings that also emerge directly from the factor array for factor 2 – see statements (#2, +3*); (#8, +5); (#37, +5*).

In terms of highlighting the added value of the audio-recorded interviews in the paper, in lines 297-301 we state that our interpretation makes use of responses to the post Q sort qualitative interviews and in line 304 that the factor solution selected was done so in part because it was consistent with the qualitative data. Examples of the added value can be seen in the italicized quotes used in our factor descriptions. For example, in Factor 2 the quote below from respondent EX009 highlights that the reasoning behind the importance of individual choice is related to the belief that the NHS should be compassionate.

“I think that if a system such as the NHS is to be truly compassionate, the patient choice and family choice has to be one of the premier things that we consider” (EX009).

Furthermore the quotes from EX029 and EX038, in conjunction with this factor 2’s factor array, highlights that life can be valuable for somebody who is dying as it can give them extra time to fulfil tasks that are important to them.

“extending life for people with terminal illnesses is only postponing death? Well I would say that my feelings again are the reverse of that, that we're enhancing what life is left” (EX029).

“if (someone wants) to achieve completion, resolution and closure around certain issues, I believe that they have a moral case for life-prolonging drugs” (EX038).

However we accept that we could make the added value gained from the audio-recorded interviews clearer and so we have added a line to the paragraph prior to the factor descriptions to reiterate that quotes taken from the short post Q sort interviews are used to enhance the factor interpretations (line 326-327) and to the methods section.

**Level of interest:** An article of importance in its field

**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