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

<table>
<thead>
<tr>
<th>TITLE (PROVISIONAL)</th>
<th>How long do patients with chronic disease expect to live? A systematic review of the literature</th>
</tr>
</thead>
<tbody>
<tr>
<td>AUTHORS</td>
<td>Hole, Barnaby; Salem, Joseph</td>
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VERSION 1 - REVIEW

<table>
<thead>
<tr>
<th>REVIEWER</th>
<th>Lucy Doos</th>
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<tr>
<td></td>
<td>Institute of Applied Health Research</td>
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<td></td>
<td>University of Birmingham</td>
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<td>UK</td>
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<tr>
<td>REVIEW RETURNED</td>
<td>06-May-2016</td>
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<table>
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<tr>
<th>GENERAL COMMENTS</th>
<th>This is a well written review that discusses an important subject which hasn't been investigated before from this prospective. However, I have got some comments.</th>
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Overall, this is an interesting and useful systematic review of studies on self-estimated life expectancy by individuals with non-cancer life-limiting illnesses. I have some comments to improve the clarity of the study and some minor remarks.

Main comments:
- The introduction is quite short and the study is poorly introduced in the last sentence, without mentioning neither the reasons for conducting the review nor the aims.
- The use of the expression life/health expectancy is not very precise: make sure you make clear the difference between self-estimated and estimated expectancies or expectations. In the first sentence of the introduction, it is not clear which type of expectation you refer to with the term “health expectations”; besides, you always talk about “life expectancy” whilst in this case you use “health expectations” which has a precise meaning and is not the same as life expectation. You could explain in the introduction the differences between self-estimated and estimated measures and clarify the terminology you will be using in the text.
- Justify why literature predating 1985 was deemed uninformative for current practice.
- Non-cancer life limiting diseases are defined only through a list of conditions. You also add that some specific conditions (e.g., cancer, HIV/AIDS) were not included and others were missing (e.g. non-COPD lung disease). As life limiting illness represent the exposure of interest in your review, I think it is important to define it also in general terms and then list all the conditions considered.
- Presenting aggregate numbers of patients included in all studies considered in your review may be uninformative and misleading. When you report it at the beginning of the results section (i.e. 545), apart from providing numbers stratified by condition, you could add at least the range of the sample sizes, from the study including the lowest to the study including the highest number of patients. Similarly, at page 7 when you say “168 of 541 patients” or “105 of 408 interviewed patients”, it is confusing: the exclusion rate among the eligible patients can be very different across studies and providing an aggregate averaged proportion is uninformative and incorrect, either report the information separately for each study or don’t report numbers. Be consistent with this suggestion throughout all text.
- Table 1: I suggest to add a column reporting the type of life-limiting illness from which patients suffered in the study.
- I have found the use of paragraphs in the results session for the comparison of self-estimated expectancy with the other measures very clear and useful. It would be nice to start each paragraph mentioning which and how many studies have used the measure of comparison (i.e. observed survival, model-predictions of survival, clinician-estimated survival), as you did for self-estimated LE compared with model-predictions of survival.
- The discussion is rather long. The findings of the review are presented and explained clearly, however the usefulness and implications of your study are just mentioned in the conclusions. Give them more emphasis and do not introduce them only in the conclusion.
- At page 16, line 7, it is not very clear what you mean with "cultural differences"

Finally, some very minor remarks:
- References: there seems to be a problem with the brackets of references 9 and 13 at page 6, and in general with punctuation, that always precedes references at the end of the sentence while full stops or comma should come after.
- Table 1 is named table A in the caption.

**REVIEWER**
Eva DuGoff
University of Wisconsin-Madison, USA

**REVIEW RETURNED**
07-Jun-2016

**GENERAL COMMENTS**
How to deliver truly patient-centered care is an important area of research, and particularly important to chronic disease management. What kind of care patient’s want is informed by how long they expect to live. This paper adds to this discussion by documenting the disconnect between patient’s self-perceived life expectancy and clinically estimated/observed life expectancy.

The manuscript is generally well-written and the conclusion which emphasizes the importance of more research of how self-estimated life-expectancy affects behavior could be important in chronic disease management. A major limitation of the findings from this study is that it included a relatively small sample of patients from medium to low quality studies. Specific concerns are described below.

Introduction
1. The magnitude of the problem is not clear. How many people are living with advanced chronic disease and how much health care do they consume? Is there any data on the high risk procedures that these individuals undergo that could result in substantial savings?
2. It would be helpful to have a more robust discussion explaining how overestimating one’s life expectancy can lead to different health care choices.

Methods
3. Page 4, line 55: According to the author, search results for publications were limited from 1985 to 2015 as papers from before 1985 were deemed unlikely to inform understanding of current practice. How was 1985 decided to be the cutoff?
4. Page 5, line 56: Did the authors develop the validity of purpose developed assessment tool? If so, how was it developed? If not, please provide relevant citations.

Results
5. Page 6, line 22: The author said that four papers reported on a single medical disease, but only three studies are cited. Please check.

Discussion
6. Page 14, line 16: Cancer is often categorized as a chronic disease. What kind of chronic diseases are you referring to here?
7. Page 14, line 41: Are there any possible explanations for why patients with ESRF only showed a similar self-estimated life expectancy and one-year mortality compared to others? And why
8. Page 15, line 37: The last sentence of this paragraph is unclear. Do you mean “care must be taken in generalising findings”?
9. It is very surprising to see how few study subjects recall discussing life expectancy with their provider. This point would be much stronger if the authors had other studies to draw on. As noted elsewhere, the included sample is quite limited and may not be representative of the experience other people with advanced chronic diseases.
10. The authors note that there are many barriers to discussing life expectancy with patients. There has been a concerted movement in the United States of America to provide patient decision aides and shared decision making tools to address major clinical decisions (e.g., back surgery). Are the authors aware of any efforts to provide life expectancy estimates through these mechanisms? If not, the authors could recommend that this be a priority area for development.
11. Do the authors have any specific policy recommendations?

**VERSION 1 – AUTHOR RESPONSE**

Reviewer(s)' Comments to Author: (annotations)

Reviewer: 1
Reviewer Name
Lucy Doos
Institution and Country
Institute of Applied Health Research
University of Birmingham
UK

Please state any competing interests or state 'None declared':
None

Please leave your comments for the authors below

This is a well written review that discusses an important subject which hasn’t been investigated before from this prospective. However, I have got some comments.

Abstract:
Objectives: Can you please add a full stop at the end of the sentence Done
Participants: Can you please added screened to the 2356 titles Done

Introduction:
- Page 4 Line 24: Can you add examples for advanced diseases. Saying “never discussed prognosis” this needs to be clear with whom they discussed. Is there any reference to support your arguing? Sentence clarified to read “incurable malignancy”. Reference provided.
- I feel that the authors need to expand (with support from the literature) on their rational for the study and the conditions selected for the study. Introduction substantially rewritten in response to reviewer comment

Methods:
- The method section needs to have subheadings such as search strategy, inclusion and exclusion criteria...etc Further subtitles added
- Authors need to add the full name rather than abbreviations when the word appears for the first time in the text. Done
- Authors didn’t report on how they dealt with disagreement. Amended with additional sentence

Results:
- In Figure 1, the authors only illustrated the number of articles from databases but what about other sources they searched. This needs to be addressed. Figure amended to include articles included in grey literature search
- Self-estimated life expectancy Vs clinician section needs to be rewritten as it is very confusing at its current status. Paragraph revised.
- Page 13 line 52: “Two studies” Can you please add the two references for them. Done
Discussion:
Page 14 line 55: “there was evidence that individuals.....” Is there any reference to support this evidence? Added references to this section
Reviewer: 2
Reviewer Name
Benedetta Pongiglione
Institution and Country
London School of Hygiene and Tropical Medicine (LSHTM), United Kingdom
Please state any competing interests or state 'None declared':
None declared
Please leave your comments for the authors below
Overall, this is an interesting and useful systematic review of studies on self-estimated life expectancy by individuals with non-cancer life-limiting illnesses. I have some comments to improve the clarity of the study and some minor remarks.
Main comments:
- The introduction is quite short and the study is poorly introduced in the last sentence, without mentioning neither the reasons for conducting the review nor the aims. Introduction substantially revised
- The use of the expression life/health expectancy is not very precise: make sure you make clear the difference between self-estimated and estimated expectancies or expectations. In the first sentence of the introduction, it is not clear which type of expectation you refer to with the term “health expectations”; besides, you always talk about “life expectancy” whilst in this case you use “health expectations” which has a precise meaning and is not the same as life expectation. You could explain in the introduction the differences between self-estimated and estimated measures and clarify the terminology you will be using in the text. Introduction substantially revised
- Justify why literature predating 1985 was deemed uninformative for current practice. Reasoning clarified in introduction and methodology, see cover letter
- Non-cancer life limiting diseases are defined only through a list of conditions. You also add that some specific conditions (e.g., cancer, HIV/AIDS) were not included and others were missing (e.g. non-COPD lung disease). As life limiting illness represent the exposure of interest in your review, I think it is important to define it also in general terms and then list all the conditions considered. Paragraph re-written to improve clarity
- Presenting aggregate numbers of patients included in all studies considered in your review may be uninformative and misleading. When you report it at the beginning of the results section (i.e. 545), apart from providing numbers stratified by condition, you could add at least the range of the sample sizes, from the study including the lowest to the study including the highest number of patients. Similarly, at page 7 when you say “168 of 541 patients” or “105 of 408 interviewed patients”, it is confusing: the exclusion rate among the eligible patients can be very different across studies and providing an aggregate averaged proportion is uninformative and incorrect, either report the information separately for each study or don’t report numbers. Be consistent with this suggestion throughout all text. Changes made in line with these comments, tracked in document
- Table 1: I suggest to add a column reporting the type of life-limiting illness from which patients suffered in the study. Done
- I have found the use of paragraphs in the results session for the comparison of self-estimated expectancy with the other measures very clear and useful. It would be nice to start each paragraph mentioning which and how many studies have used the measure of comparison (i.e. observed survival, model-predictions of survival, clinician-estimated survival), as you did for self-estimated LE compared with model-predictions of survival. Done
- The discussion is rather long. The findings of the review are presented and explained clearly,
however the usefulness and implications of your study are just mentioned in the conclusions. Give them more emphasis and do not introduce them only in the conclusion. Changes made to discussion and conclusion in line with these and other reviewers’ comments
- At page 16, line 7, it is not very clear what you mean with “cultural differences” Rephrased
Finally, some very minor remarks:
- References: there seems to be a problem with the brackets of references 9 and 13 at page 6, and in general with punctuation, that always precedes references at the end of the sentence while full stops or comma should come after. References amended
- Table 1 is named table A in the caption. Amended

Reviewer: 3
Reviewer Name
Eva DuGoff
Institution and Country
University of Wisconsin-Madison, USA
Please state any competing interests or state ‘None declared’:
None Declared
Please leave your comments for the authors below
How to deliver truly patient-centered care is an important area of research, and particularly important to chronic disease management. What kind of care patient’s want is informed by how long they expect to live. This paper adds to this discussion by documenting the disconnect between patient’s self-perceived life expectancy and clinically estimated/observed life expectancy.
The manuscript is generally well-written and the conclusion which emphasizes the importance of more research of how self-estimated life-expectancy affects behavior could be important in chronic disease management. A major limitation of the findings from this study is that it included a relatively small sample of patients from medium to low quality studies. Specific concerns are described below.

Introduction
1. The magnitude of the problem is not clear. How many people are living with advanced chronic disease and how much health care do they consume? Is there any data on the high risk procedures that these individuals undergo that could result in substantial savings? Introduction substantially rewritten
2. It would be helpful to have a more robust discussion explaining how overestimating one’s life expectancy can lead to different health care choices. Introduction substantially rewritten

Methods
3. Page 4, line 55: According to the author, search results for publications were limited from 1985 to 2015 as papers from before 1985 were deemed unlikely to inform understanding of current practice. How was 1985 decided to be the cutoff? Reasoning clarified in introduction and methodology, see cover letter.
4. Page 5, line 56: Did the authors develop the validity of purpose developed assessment tool? If so, how was it developed? If not, please provide relevant citations. Paragraph amended

Results
5. Page 6, line 22: The author said that four papers reported on a single medical disease, but only three studies are cited. Please check. Done

Discussion
6. Page 14, line 16: Cancer is often categorized as a chronic disease. What kind of chronic diseases are you referring to here? Amended
7. Page 14, line 41: Are there any possible explanations for why patients with ESRF only showed a similar self-estimated life expectancy and one-year mortality compared to others? And why did it reversed after two years? Sentence added
8. Page 15, line 37: The last sentence of this paragraph is unclear. Do you mean “care must be taken in generalising findings” Sentence amended
9. It is very surprising to see how few study subjects recall discussing life expectancy with their provider. This point would be much stronger if the authors had other studies to draw on. As noted
elsewhere, the included sample is quite limited and may not be representative of the experience other people with advanced chronic diseases. Substantial amendment made to discussion

10. The authors note that there are many barriers to discussing life expectancy with patients. There has been a concerted movement in the United States of America to provide patient decision aides and shared decision making tools to address major clinical decisions (e.g., back surgery). Are the authors aware of any efforts to provide life expectancy estimates through these mechanisms? If not, the authors could recommend that this be a priority area for development. Comments added to conclusion

11. Do the authors have any specific policy recommendations? Conclusion amended

(end of comments)


VERSION 2 – REVIEW

<table>
<thead>
<tr>
<th>REVIEWER</th>
<th>Benedetta Pongiglione</th>
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<tbody>
<tr>
<td></td>
<td>London School of Hygiene and Tropical Medicine, London UK</td>
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| REVIEW RETURNED | 15-Aug-2016 |

**GENERAL COMMENTS**
The introduction is now more definite and the rational for the study and the aims more clearly illustrated. The first paragraph presents quite detailed statistics of chronic disease in the UK. I understand this addresses one of the reviewers’ comments asking to clarify the magnitude of the problem of chronic disease, but I have found that starting with a list of numbers before elucidating the background and rationale of the study is not very engaging for the reader and ends up being a bit distracting.

A few minor remarks:

- Line 16-18 page 5: the sentence “In the UK...including intensive care” seems a bit disconnected and unnecessary, drop it if you can.
- Line 54 page 14: paragraph title, add “life expectancy”, such that it becomes “self-estimated life expectancy compared with clinician-estimated life expectancy”
- Table 1: thank you for addressing my comment and add the conditions to the table. However, I think it would be clearer not to put it in the same cell with author's name. If you are short of space do rather combine together author's name and year of publication, and use a separate column for condition.
This is the second round revision of the manuscript. The authors have corrected most issues identified in the first round.

I continue to be concerned that 1985 is an arbitrary cut-off. The decision to limit studies to those after 195 may introduce some bias though the extent may vary by disease.

**Reviewer: 1**

This is the second round revision of the manuscript. The authors have corrected most issues identified in the first round. I continue to be concerned that 1985 is an arbitrary cut-off. The decision to limit studies to those after 195 may introduce some bias though the extent may vary by disease.

Response – search updated as detailed above

**Reviewer: 2**

The introduction is now more definite and the rational for the study and the aims more clearly illustrated. The first paragraph presents quite detailed statistics of chronic disease in the UK. I understand this addresses one of the reviewers’ comments asking to clarify the magnitude of the problem of chronic disease, but I have found that starting with a list of numbers before elucidating the background and rationale of the study is not very engaging for the reader and ends up being a bit distracting.

Response – introduction revised and simplified to reflect these comments.

A few minor remarks:

Line 16-18 page 5: the sentence “In the UK…including intensive care” seems a bit disconnected and unnecessary, drop it if you can.

Response – sentence removed.

Line 54 page 14: paragraph title, add “life expectancy”, such that it becomes “self-estimated life expectancy compared with clinician-estimated life expectancy”

Response – done.

Table 1: thank you for addressing my comment and add the conditions to the table. However, I think it would be clearer not to put it in the same cell with author’s name. If you are short of space do rather combine together author’s name and year of publication, and use a separate column for condition.

Response – done, as suggested with author/date and condition in columns.
How long do patients with chronic disease expect to live? A systematic review of the literature

Barnaby Hole and Joseph Salem

*BMJ Open* 2016 6:
doi: 10.1136/bmjopen-2016-012248

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