Author’s response to reviews

Title: Prostate Cancer Survivors Preferences on the Delivery of Diet and Lifestyle Advice. A Pilot Best-Worst Discrete Choice Experiment

Authors:

Luke Robles (luke.robles@bristol.ac.uk)
Stuart J Wright (stuart.wright-2@manchester.ac.uk)
Lucy Hackshaw-McGeagh (lucy.hackshaw@bristol.ac.uk)
Ellie Shingler (ellie.shingler@bristol.ac.uk)
Constance Shiridzinomwa (constance.shiridzinomwa@nbt.nhs.uk)
J. Athene Lane (athene.lane@bristol.ac.uk)
Richard M Martin (richard.martin@bristol.ac.uk)
Sorrel Burden (sorrel.burden@manchester.ac.uk)

Version: 1 Date: 17 Jun 2019

Author’s response to reviews:

4th June 2019

Dear Editors,

Thank you for your email on 7th May 2019. We are grateful for the opportunity to revise and resubmit our manuscript. We thank the reviewers for their helpful comments.

Please find below a point-by-point response to the reviewers’ comments. We have highlighted the amendments to the manuscript with red text. For ease of reference, we have included the additional text to the manuscript in response to the reviewers’ comments below where appropriate.

Reviewer #1 comments:

1. The authors should consider adding in some additional details on study design and population to the abstract, such as the age group. The authors state that this is a cross-sectional pilot study which was nested within a feasibility RCT. This would be useful information to have in the abstract.
Response: As requested, we amended the abstract to clearly state the study design and age of participants. Please see lines 52-54.

2. In the introduction the authors discuss the use of econometric analysis and an out-of-pocket cost attribute (Pg. 5; Ln. 104-106). Some additional information and references on these would be of use to readers.

Response: Econometric analysis is a broad term to cover a range of mainly regression-based approaches to analyse economic data. We agree that a further explanation on both this analysis and the out-of-pocket cost attribute is required. To avoid confusion, we have removed the terms, econometric analysis, from the text and have elaborated on how the discrete choice experiment was administered and analysed and included two additional references in lines 103-107:

Participants are shown questions containing a number of scenarios, which are created by combining these attributes and levels, and are traditionally asked to choose their most preferred scenario. This produces a binary outcome, whether a particular scenario was chosen or not, which means that logistic regression or probit models can be used to determine which attributes and levels were most likely to predict the chosen scenario [16,17]. We have also provided a more detailed description of the out-of-pocket attribute and have provided a reference for this in lines 107-113:

An out-of-pocket cost attribute can be included to represent the inconvenience that participants would experience in accessing the service. For example, patients may have to forgo work or leisure activities in order to attend the health service and this has a cost to them. The inclusion of such attributes allows the calculation of the willingness to pay for attributes and levels. These figures represent the maximum inconvenience (in terms of cost) that participants would be willing to go through to receive beneficial aspects of the service and, therefore, may provide a more intuitive method for valuing the levels of the service than the coefficients of the logistic regression model.

3. The authors state that the participants were diagnosed with localised (low risk) prostate cancer (Pg. 5; Ln. 127). Is there a standardised definition for this?

Response: Localised prostate cancer is the clinical description of prostate cancer that has not spread outside the prostate gland. We realised that this description may be confusing for non-clinical audiences. Therefore, we have amended the respective sentence (line 134-135) to clarify what we mean by localised prostate cancer:

Participants were men aged 18 or over, who were diagnosed with localised (low risk) prostate cancer (i.e. cancer that has not spread outside the prostate gland) within the last 12 months, and who had undergone radical prostatectomy.

4. The authors use the wording "attribute is no longer attractive" (Pg. 6; Ln. 144). Could the authors explain this more or standardise the wording against what is used in the results/discussion sections.

Response: This wording refers to the out-of-pocket costs and whether the cost of receiving the information outweighs the benefits received from the service. We agree that this wording is unclear and requires further explanation. To avoid confusion, we have removed 'attribute is no
longer attractive’ and provided a more detailed description of how we decided on the appropriate level of out-of-pocket costs in lines 150-157:

This is achieved by calculating the amount of additional cost that men are willing to incur for a service featuring a specific attribute or level before the cost outweighs the benefit they would receive from the service. These monetary values show how valuable each attribute is to the population. The appropriate level of out-of-pocket costs to force participants to trade was determined in the Best-Worst DCE reported by Wright et al., [21] on which this study was based. In the initial pilot study for that DCE, participants did not use the cost in their decision making, indicating the values were not enough to outweigh the benefits of the health service. Following the doubling of the values for the main study, cost became a significant factor in participants’ choices.

5. The authors state that they "create a set of questions using a d-efficient design created in NGene" (Pg. 6; Ln. 147), could this be explained in more detail?

Response: An efficient design is used to construct the choice questions to ensure that all levels appear in the questions asked to allow for their coefficients to be estimated. The design also ensures that these coefficients can be estimated with the minimum possible error. We have amended the respective paragraph with the following sentences in lines 161-163 to make this clearer:

This means that the error in measuring preferences introduced by the questionnaire is minimised, thereby improving the coefficient estimates. This also ensures that the coefficients of all attributes can be estimated unlike in random designs where some levels may accidentally not be included.

6. The authors use the wording "simplifying heuristics" (Pg. 7; Ln. 176), and "heteroscedastic" (Pg. 8; Ln. 183), could this be explained in more detail?

Response: We agree that further detail is required on what we mean by these terms. We have amended the statistical analysis section regarding ‘simplifying heuristics’ with the following paragraph in lines 191-196:

There is a lot of information to take on in each DCE question and participants may make choices based on only a few attributes or by other rules such as never choosing profiles containing a certain level. These approaches are known as simplifying heuristics and it has been argued that if preferences are significantly different for best and worst choices then participants may be using such heuristics to make choices rather than fully evaluating all attributes and levels. In such situations if may be inappropriate to combine the best and worst data due to a violation of the underlying assumptions of the choice model.

We have also amended the paragraph in lines 201-204 to clarify what we mean by heteroscedastic:

Such differences, known as scale heterogeneity or heteroscedasticity, may result in different levels of error variance in the responses to the best and worst questions which can bias the results of combined models. As such a heteroscedastic best-worst logistic regression was used to test for statistically significant differences in scale.
7. The authors talk about "econometric analysis" in the introduction, was this type of analysis conducted?

Response: As per comment 2 above, econometric analysis is a broad term to cover a range of mainly regression-based approaches to analyse economic data. To avoid confusion, we have removed the terms, econometric analysis, from the text and have elaborated on how the discrete choice experiment was administered and analysed. Please see lines 103-113 in the manuscript.

General comments

1. Standardise the use of follow-up versus follow up

2. Standardise the use of numbers versus words (i.e. 6 versus six)

Response: Thank you for these helpful comments. We have amended the text for consistency and have used ‘follow-up’ (hyphenated) and numerical digits for numbers instead of words.

Reviewer #2: This is a well-written and worthwhile study. There are a few suggestions to improve the manuscript for readers:

-Clarifying whether each questionnaire included the same 12 scenarios and providing further information on how these were exactly selected.
Response: Each participant was presented with the same 12 scenarios. We have clarified this in line 145:
‘The BWDCE questionnaire presented each participant with a set of the same 12 hypothetical scenarios.’

We have also added more detail on how the questions were selected in lines 158-163:
‘Each attribute contained four levels which described how or when information is given and by whom. These levels were then combined to create a set of 12 questions using a d-efficient design created in NGene software [22] to improve the statistical efficiency of the collected data by minimising the generalised variance of the parameter estimates. This means that the error in measuring preferences introduced by the questionnaire is minimised, thereby improving the coefficient estimates. This also ensures that the coefficients of all attributes can be estimated unlike in random designs where some levels may accidentally not be included.’

-Providing further information on how out of pocket/indirect cost of receiving information were determined for the scenarios.
Response: We provided a more detailed description of the out-of-pocket attribute and have provided a reference for this. We have changed the text in accordance with this comment in lines 103-113:

Participants are shown questions containing a number of scenarios, which are created by combining these attributes and levels, and are traditionally asked to choose their most preferred scenario. This produces a binary outcome, whether a particular scenario was chosen or not, which means that logistic regression or probit models can be used to determine which attributes
and levels were most likely to predict the chosen scenario [16,17]. An out-of-pocket cost attribute can be included to represent the inconvenience that participants would experience in accessing the service. For example, patients may have to forgo work or leisure activities in order to attend the health service and this has a cost to them. The inclusion of such attributes allows the calculation of the willingness to pay for attributes and levels. These figures represent the maximum inconvenience (in terms of cost) that participants would be willing to go through to receive beneficial aspects of the service and, therefore, may provide a more intuitive method for valuing the levels of the service than the coefficients of the logistic regression model.

- What were the response options for the 3 questions: 1) how difficult they found making choices; 2) whether they used all attributes in their decision; and 3) which attribute they found most important in their decision?

Response: We have now included the response options to the above questions in lines 168-172: These questions asked: 1) how difficult they found making choices (very easy, easy, neither easy or hard, hard, very hard); 2) which attributes they used in their decision (how the information is given, where the information is given, who gives the information, cost, all of these); and 3) which attribute they found most important in their decision (how the information is given, where the information is given, who gives the information, cost, all of these).

-Further discussion on the point 'further work might need to evaluate the reliability of the BWDCE'

Response: We have elaborated on what we mean by further work in the discussion section in lines 321-324: However, further work might need to evaluate the reliability of the BWDCE in evaluating men’s preferences. A larger study may be necessary to ensure the preferences of the men in this study are representative of those in the wider population of prostate cancer survivors. Further qualitative interviews may be also useful to determine the acceptability of the results in guiding clinical practice.

-Discussion regarding the implications of sample size on their findings. Could it have an impact on whether a level of an attribute reached statistical significance or not?

Response: This is an interesting question with regards to best-worst DCEs and the additional data elicited from participants means that statistical efficiency can be achieved at a relatively small sample size. However, this can be misleading as the pilot study sample is not representative of the wider population. It may therefore be that increasing the sample size for such studies actually increase heterogeneity. We have amended the limitations section to try to represent this interesting aspect of the study design in lines 309-314: If the preferences of this larger sample are similar to that of the study population then the coefficients will be estimated with greater accuracy. However, this may introduce a more heterogeneous population that does not increase the statistical significance of the coefficients due to preference heterogeneity. In this case, the use of latent class analysis, such as that reported in this study, would become more reliable and critical in order to provide evidence as to how the intervention could be tailored to best meet patients’ preferences.

Minor typo:
We conducted a pilot best-worst discrete choice experiment questionnaire to explore how men's preferences of receiving diet and physical activity advice following surgery for localised prostate cancer...remove 'how'

Response: Thank you for identifying this typo. We have amended this sentence in line 53 in accordance with the comment above.

Yours faithfully,

Dr Sorrel Burden