VALUING HEALTH AT THE END OF LIFE: AN EXAMINATION OF FRAMING EFFECTS AND STUDY DESIGN CONSIDERATIONS

REPORT BY THE DECISION SUPPORT UNIT

[September 2019]

Koonal K. Shah[1,2,3], Aki Tsuchiya[2,4], Allan J. Wailoo[2]

[1] Office of Health Economics

[2] School of Health and Related Research, University of Sheffield

[3] PHMR Ltd

[4] Department of Economics, University of Sheffield

Decision Support Unit, ScHARR, University of Sheffield, Regent Court, 30 Regent Street
Sheffield, S1 4DA

Tel (+44) (0)114 222 0734
E-mail dsuadmin@sheffield.ac.uk
Website www.nicedsu.org.uk
Twitter @NICE_DSU

ABOUT THE DECISION SUPPORT UNIT

The Decision Support Unit (DSU) External Assessment Centre is based at the University of Sheffield with members at York, Bristol, Leicester and the London School of Hygiene and Tropical Medicine. The DSU is commissioned by The National Institute for Health and Care Excellence (NICE) to provide a research and training resource to support the Institute's Centre for Health Technology Evaluation Programmes. Please see our website for further information www.nicedsu.org.uk.

The production of this document was funded by the National Institute for Health and Care Excellence (NICE) through its Decision Support Unit. The views, and any errors or omissions, expressed in this document are of the authors only. NICE may take account of part or all of this document if it considers it appropriate, but it is not bound to do so.

This report should be referenced as follows:

Shah, K.K., Tsuchiya, A. and Wailoo, A.J., Valuing health at the end of life: an examination of framing effects and study design considerations. NICE DSU Report 2019.

EXECUTIVE SUMMARY

Objective: A number of recent studies have examined the extent of public support for an ‘end of life premium’ – that is, whether people place greater weight on a unit of health gain for end of life patients than on that for other types of patients. The objective of this study is to assess whether any observed preferences regarding an end of life premium are affected by framing effects and study design considerations, such as the perspective used to elicit preferences and whether or not visual aids and indifference options are included in the survey.

Methods: Preferences were elicited from a representative sample of the UK general public using an online survey (n=2401). Respondents were randomly allocated to one of six study arms, each of which applied a different framing. The study design was informed by the National Institute for Health and Care Excellence's supplementary policy appraising life-extending end of life treatments. The choice tasks involved asking respondents which of two hypothetical patients they would prefer to treat, assuming there were enough funds to treat only one of them. Respondents were also asked a series of attitudinal questions examining their support for general health care priority setting policies. Comparisons between arms and between tasks were assessed using the Pearson’s chi-squared test.

Results: The overall results were not consistent with an end of life premium. Respondents’ choices were found to be sensitive to the choice of perspective, and to the inclusion of indifference options and (to a lesser extent) visual aids. However, in none of the study arms did a majority of respondents choose to prioritise the treatment of the end of life patient.

Conclusions: The findings demonstrate the influence of framing effects and study design considerations in stated preference research. Researchers should seek to control for such effects when seeking to examine people’s health care priority setting preferences.

KEYWORDS

End of life; NICE; stated preferences; public preferences; social preferences; framing effects; indifference

CONTENTS

1. INTRODUCTION ... 7
2. METHODS ... 10
  2.1. SURVEY INSTRUMENT ... 10
    2.1.1. Scenarios S1 to S7 ... 10
    2.1.2. Follow-up questions for scenarios S1 to S7 ... 14
    2.1.3. Debrief questions regarding scenarios S1 to S7 ... 15
    2.1.4. Scenario S8 ... 15
    2.1.5. Attitudinal questions ... 16
    2.1.6. Study design ... 17
  2.2. ADMINISTRATION OF SURVEY ... 20
  2.3. SAMPLE ... 20
  2.4. ETHICAL APPROVAL ... 21
  2.5. PILOTING ... 21
    2.5.1. Summary of responses to the debrief questions ... 22
    2.5.2. Improvements made as a result of the pilot ... 24
  2.6. METHODS OF ANALYSIS ... 25
3. RESULTS ... 28
  3.1. AGGREGATE RESPONSES TO SCENARIO QUESTIONS ... 31
  3.2. IMPACT OF EXPERIMENTAL MODIFICATIONS ... 34
    3.2.1. Impact of including an indifference option ... 34
    3.2.2. Impact of including a visual aid ... 35
  3.3. RESPONSES TO FOLLOW-UP QUESTIONS ... 35
  3.4. SETS OF CHOICES ... 37
  3.5. CROSS-TABULATIONS OF RESPONSES FROM SELECTED PAIRS OF SCENARIOS ... 39
  3.6. DEBRIEF STATEMENTS ... 42
  3.7. SENSITIVITY ANALYSIS: EXCLUSIONS LINKED TO DATA QUALITY ... 44
  3.8. ATTITUDINAL QUESTIONS ... 45
  3.9. IMPACT OF RESPONDENT BACKGROUND CHARACTERISTICS IN S1 ... 48
  3.10. ORDERING EFFECTS ... 49
4. DISCUSSION ... 52
5. CONCLUSIONS ... 62
6. REFERENCES ... 63

TABLES

Table 1. Summary of scenarios S1 to S7 ... 11
Table 2. Study arms and survey versions ... 18
Table 3. Survey completion and exclusion statistics ... 28
Table 4. Time taken (in minutes) to complete survey ... 28
Table 5. Sample background characteristics ... 30
Table 6. S1: End of life patient (A) vs. non-end of life patient (B) ... 31
Table 7. S2: Older end of life patient (A) vs. younger non-end of life patient (B) ... 31
Table 8. S3: End of life patient with more time with knowledge (A) vs. non-end of life patient with less time with knowledge (B) ... 32
Table 9. S4: Quality of life improvement for end of life patient (A) vs. life extension for end of life patient (B) ... 32
Table 10. S5: Quality of life improvement for end of life patient (A) vs. life extension for non-end of life patient (B) ... 32

Table 11. S6: Quality of life improvement at end of normal life expectancy (A) vs. life extension at end of normal life expectancy (B) 33
Table 12. S7: Smaller life extension for end of life patient (A) vs. larger life extension for end of life patient (B) 33
Table 13. S8: Individual perspective adaptation of S1 – 1 year of life expectancy without treatment (A) vs. 5 years of life expectancy without treatment (B) 34
Table 14. Summary of responses to follow-up questions, by scenario 36
Table 15. Five most common sets of choices in S1 to S7 38
Table 16. Selected statistics for respondents expressing indifference in all scenarios (III-III-I) and respondents who choices reflect QALY-maximisation (III-III-B) 39
Table 17. Cross-tabulation – S1 vs. S2 40
Table 18. Cross-tabulation – S1 vs. S3 40
Table 19. Cross-tabulation – S1 vs. S5 40
Table 20. Cross-tabulation – S4 vs. S6 41
Table 21. Cross-tabulation – S1 vs. S8 41
Table 22. Cross-tabulation – S1 vs. S8 (survey version 5 only) 42
Table 23. S1 choices made by all respondents and those who hit data quality flags 45
Table 24. S1 choices after exclusions based on flags 1 and 2 45
Table 25. Responses to attitudinal questions 46
Table 26. Responses to attitudinal questions 47
Table 27. Cross-tabulation: S4 vs. attitudinal statement 4 47
Table 28. Cross-tabulation: S4 vs. attitudinal statement 5 47
Table 29. Cross-tabulation: S4 vs. attitudinal statement 6 47
Table 30. Impact of background characteristics – results of regression 48
Table 31. Aggregate S1 to S6 responses, split by whether scenario appeared first or not 50

FIGURES

Figure 1. Screenshot from survey (S1; visual aid arm; forced choice arm) 19
Figure 2. Screenshot from survey (S1; no visual aid arm; indifference option 1 arm) 19
Figure 3. Responses to S4-S6 follow-up questions made by respondents who initially chose the quality of life improvement option in S4-S6 37
Figure 4. Responses to debrief statement 1 (by indifference arm) 43
Figure 5. Responses to debrief statement 1 (by visual aid arm) 43
Figure 7. Responses to debrief statement 2 (by visual aid arm) 44
Figure 8. Proportion choosing A or B versus proportion choosing I, by scenario order 51

1. INTRODUCTION

Public preferences have been cited as a rationale behind the way in which life-extending end of life treatments are appraised by the National Institute for Health and Care Excellence (NICE)[1]. Since 2009, NICE has applied a supplementary policy which indicates that if certain criteria are met, it may be appropriate to recommend the use of such treatments even if their cost-effectiveness estimates exceed the range normally considered acceptable[2]. However, there have been concerns that there is little evidence to support the premise that society places special value on life-extending end of life treatments. A recent review found that the evidence was mixed overall, identifying eight studies reporting evidence consistent with an end of life premium, and 11 studies reporting evidence not consistent with an end of life premium[3]. The question of whether members of the public wish to place greater weight on a unit of health gain for end of life patients than on that for other types of patients thus remains unresolved.

The review highlighted a number of gaps in the literature. Age-related preferences were not controlled for in some studies, which makes it difficult to disentangle preferences for prioritising the treatment of end of life patients from preferences for prioritising the treatment of the relatively young (or old). Aside from the authors' own studies, none of the studies reviewed attempted to control for time-related preferences. Previous research by the authors suggests that the preference for prioritising the treatment of end of life patients (where observed) may be driven by concern about how little time those patients have known about their prognosis, and therefore how little time they have to prepare for death[4-6]. The issue of preparedness has generated interest amongst academic and industry audiences during presentations and discussions of the authors' research (Cowell, W., 2013, personal communication, 26 March; Longworth, L., 2013, personal communication, 26 March; McHugh, N., 2013, personal communication, 26 March). However, with the exception of Cookson[7] and McHugh et al.[8], it has received limited attention in the literature to date.

A further gap in the literature is that few studies tested the robustness of their results – for example, by checking whether respondents agreed with researchers' interpretations of their responses to the choice tasks; or by checking whether consistent results could be obtained using different study designs or methodologies. The review provided some evidence that the results of the empirical studies may have been influenced by the choice of method and the way in which the choice tasks were framed and operationalised. For example, studies that included visual aids and/or indifference options, and studies instructing respondents to adopt an individual/own health perspective (as opposed to a social decision-maker perspective) appear more likely than average to report evidence consistent with an end of life premium. However, the small number of studies in the sample makes it difficult to make conclusive claims about the existence of such effects.

The choice of perspective to be used when eliciting health care priority-setting preferences has been discussed in the literature[9, 10]. It should be noted that all of the individual perspective studies included in the literature review used the willingness-to-pay method, which may have been a more influential factor than the choice of study perspective per se.

The aim of this study is to add to the literatures on people's preferences regarding health care priority-setting (in particular, regarding the prioritisation of the treatment of end of life patients) and on framing effects in stated preference research.
Specific objectives are to test the following null hypotheses:

1. People place no more weight on a unit of health gain for end of life patients than on that for other types of patients, ceteris paribus.
2. Any observed preferences regarding an end of life premium are unaffected by whether or not the end of life patient is older than the non-end of life patient.
3. Any observed preferences regarding an end of life premium are unaffected by whether or not the end of life patient has known about their prognosis for longer than the non-end of life patient.
4. People place no more weight on life-extending treatments than on quality of life-improving treatments for end of life patients, controlling for the size of the gain.
5. Any observed preferences regarding an end of life premium are unaffected by whether the end of life treatment is quality of life-improving or life-extending.

6. Any observed preferences between quality of life improvements and life extensions are unaffected by whether the gains occur in an end of life or a non-end of life context.
7. Any observed preferences regarding an end of life premium are unaffected by whether the preferences are being elicited from an individual or a social decision-maker perspective.
8. Any observed preferences regarding an end of life premium are unaffected by whether visual aids are included in the stated preference survey.
9. Any observed preferences regarding an end of life premium are unaffected by whether an indifference option is included (or by the wording of the indifference option) in the stated preference survey.

A further objective is to examine the consistency of people's views by using two different approaches (choice exercise and attitudinal statements with Likert item responses) to infer their preferences in relation to the hypotheses above.

---

[1] Following Plous[11], framing effects are defined as an example of cognitive bias whereby people's reaction to a given choice is influenced by the way is which that choice is presented – for example, using visual or non-visual presentation. Framing is considered problematic in stated preference research because it results in respondents making choices using irrelevant information that is not intended to convey information about the value of the choice options[12]. Framing effects can be distinguished from the effects of study design choices that are intended to provide relevant information to respondents, such as the choice of study perspective.

2. METHODS

2.1. SURVEY INSTRUMENT

A self-completion internet survey was developed in collaboration with epiGenesys, a software development company. The same company was commissioned to help develop the survey used in Shah *et al*.[6]. The survey comprised the following elements (in order):

- Background / screening questions
- Information sheet and consent form
- Instructions (including explanation of the diagrams, if relevant)
- Seven scenarios (S1 to S7) requiring respondents to adopt a social decision-maker perspective
- Two debrief questions (Likert items)
- One scenario (S8) requiring respondents to adopt an individual perspective
- Six attitudinal questions (Likert items)
- Further background questions

The primary method used in this study was a choice exercise similar to that used by Shah *et al*.[5]. The DCE approach was not used in this study. This method is useful when it is desirable to examine multiple levels for a small number of attributes and to predict preferences over scenarios that are not actually presented, but it is less suitable when testing hypotheses regarding the isolated impact of a large number of attributes and study design approaches.

2.1.1. Scenarios S1 to S7

Following the approach used by Shah *et al.*[5, 6], each scenario presented information about two hypothetical individuals (patient A and patient B). Both patients could benefit from treatment, and the respondents were asked to assume that the health service had enough funds to treat one but not both of them. The patients and their circumstances were described in terms of the following attributes:

- Age today (years)
- Age at death without treatment (years)
- Timing of diagnosis (the patients were described either as having 'just been diagnosed' or as having been 'diagnosed 5 years ago')
- Life expectancy without treatment (from today) (years)
- Quality of life without treatment (%)[1]

---

[2] Note that, as with the authors' previous empirical studies, the terms 'health' and 'general health' (distinct from life expectancy) were presented to respondents rather than 'quality of life'. In what follows, the term 'quality of life' is used unless specifically referring to the wording of the survey or when the more general meaning of the term 'health' (encompassing both quality of life and length of life) is intended.

- Gain from treatment (months or %, depending on whether the gain was a life extension or a quality of life improvement, respectively)

The initial question in each scenario required respondents to adopt the perspective of a social decision-maker and to indicate which of the following statements best described their view: (1) I would prefer the health service to treat patient A; or (2) I would prefer the health service to treat patient B. Some of the respondents were also offered a third option whereby they could express indifference between treating patient A and patient B (see 2.1.6).

Table 1 summarises the information provided to the respondents for scenarios S1 to S7.

Scenario S1 tests whether respondents wish to give priority to the end of life patient (patient A, whose life expectancy of one year without treatment meets the NICE criterion for defining ‘short life expectancy’) or to the non-end of life patient (patient B, whose life expectancy of five years does not meet the NICE criterion). The only other difference between the two patients is that patient A is described as being four years older than patient B today, though both patients would die at the same age without treatment. A preference for treating patient A can be interpreted as evidence consistent with an end of life premium (hypothesis 1).

Table 1. Summary of scenarios S1 to S7

	S1	S2	S3	S4	S5	S6	S7
Age today							
Patient A	49 years	69 years	49 years	49 years	49 years	50 years	49 years
Patient B	45 years	45 years	45 years	49 years	45 years	50 years	49 years
Age at death without treatment							
Patient A	50 years	70 years	50 years	50 years	50 years	80 years	50 years
Patient B	50 years	50 years	50 years	50 years	50 years	80 years	50 years
Timing of diagnosis[a]							
Patient A	JD	JD	5Y	JD	JD	JD	JD
Patient B	JD	JD	JD	JD	JD	JD	JD
Life expectancy without treatment							
Patient A	1 year	1 year	1 year	1 year	1 year	30 years	1 year
Patient B	5 years	5 years	5 years	1 year	5 years	30 years	1 year
Quality of life without treatment							
Patient A	100%	100%	100%	50%	50%	50%	100%
Patient B	100%	100%	100%	50%	50%	50%	100%
Life expectancy gain from treatment							
Patient A	+12 mths	+12 mths	+12 mths	None	None	None	+6 mths
Patient B	+12 mths	+12 mths	+12 mths	+12 mths	+12 mths	+12 mths	+12 mths
Quality of life gain from treatment							
Patient A	None	None	None	+50%	+50%	+50%[c]	None
Patient B	None	None	None	None	None	None	None
Undiscounted QALY gain from treatment[b]							
Patient A	1 QALY	1 QALY	1 QALY	0.5 QALY	0.5 QALY	0.5 QALY	0.5 QALY
Patient B	1 QALY	1 QALY	1 QALY	0.5 QALY	0.5 QALY	0.5 QALY	1 QALY

[a] JD: just been diagnosed; 5Y: diagnosed five years ago
[b] Respondents did not see this information (the term ‘QALY’ was not used at any point in the survey)
[c] Quality of life gain achieved in final year of life only

In order to examine whether any observed preference for treating end of life patients over non-end of life patients is driven by the relative current ages of the patients, scenario S2 replicates S1 except that patient A is 69 years today (rather than 49 years) and would die aged 70 years without treatment (rather than 50 years). If respondents switch from preferring to treat patient A in S1 to preferring to treat patient B in S2, this can be interpreted as evidence that the preference for treating the end of life patient depends on the age of the patient (hypothesis 2).

In order to examine whether any observed preference for treating end of life patients over non-end of life patients is driven by how long the patients have known about their prognosis, scenario S3 replicates S1 except that patient B’s illness is described as having been diagnosed five years ago (rather than having just been diagnosed). Patient A’s illness is described as having just been diagnosed in both S1 and S3. Hence, while patient A’s life expectancy without treatment is shorter than that of patient B, patient A has known about, and (by their expected time of death) will have known about, their prognosis for longer than patient B. If respondents switch from preferring to treat patient A in S1 to preferring to treat patient B in S3, this can be interpreted as evidence that the preference for treating the end of life patient depends on how long the patient has known about their prognosis (hypothesis 3).

In scenarios S1 to S3, the patients’ illnesses were described as affecting their life expectancy but not their general level of health (as noted above, the terms ‘health’ and ‘general health’ were used in the survey to distinguish quality of life from length of life), and the treatments available were described as offering life extensions but would not affect their general level of health. In scenario S4, on the other hand, both patients are described as experiencing a poorer level of health as a result of their illnesses – they would be in ‘50% health’ without treatment. The treatment for patient A would restore them to full health (with no effect on life expectancy), whereas the treatment for patient B would extend their life by 12 months (with no effect on general health). An observed preference for treating patient A or patient B therefore indicates whether people wish to give higher priority to quality of life-improving or life-extending treatments for end of life patients, respectively (hypothesis 4).

Scenario S5 replicates S4 except that patient B is now described as being 45 years old today (rather than 49 years old) and has a life expectancy of five years without treatment (rather than one year). The choice in this scenario is therefore between a quality of life improvement for an end of life patient and a life extension for a non-end of life patient. If respondents switch from preferring to treat patient A in S1 to preferring to treat patient B in S5, this can be interpreted as evidence that the preference for treating the end of life patient relies on the treatment for the end of life patient being life-extending (hypothesis 5).

The findings reported by Shah *et al*.[5] suggest that the majority of respondents will choose to treat patient A in S4. This would imply that people believe that quality of life-improving end of life treatments should be prioritised over life-extending end of life treatments. In the instructions, the concept of ’50% health’ was explained as follows:

> Suppose there is a health state which involves some health problems. If patients tell us that being in this health state for 2 years is equally desirable as being in full health for 1 year, then we would describe someone in this health state as being in 50% health.

Based on such an assumption, a 50% quality of life improvement (lasting 12 months) can be said to be generate gains for patient A that are equal in size to the gains for patient B generated by a 12-month life extension (at 50% quality of life). If respondents still express a preference for treating patient A, this suggests that they consider the quality of life improvement to be more *socially* valuable than the life extension, at least in the end of life context where both patients have one year left to live. The purpose of scenario S6 was to test whether quality of life improvements or life extensions were preferred in a *non-end of life* context. Depending on whether respondents make the same or different choices in S4 and S6, the results could imply either that the preferences observed in S4 are specific to the end of life context or that the respondents are seeking to impose a (general, non-end of life-specific) social value judgement onto the QALY model[13]. Switches in choices between S4 and S6 can be interpreted as evidence that the preference between quality of life improvements and life extensions is context-specific (hypothesis 6).

Given concerns about the quality of internet survey data[14], it is useful to include a task that can act as a ‘rationality check’, helping to identify respondents whose choices suggest a poor level of attentiveness, engagement or understanding. Scenarios S1 to S6 were designed such that both patients gained the same number of undiscounted QALYs from treatment (1 QALY in S1, S2 and S3; half a QALY in S4, S5 and S6). Scenario S7, on the other hand, involves a choice between a smaller life extension (6 months) for patient A and a larger life extension (12 months) for patient B, with all other attributes at the same level in both alternatives. A respondent who supports a QALY-maximisation objective to health care priority-setting, or indeed simply one who considers a greater number of QALYs gained to be a good thing, should in theory consider treating patient B to be more valuable than treating patient A. Even respondents who reject the notion that priority-setting decisions should be guided by information about the size of the QALY gains should (in theory) be indifferent between treating patient A and patient B. Hence, treating patient A can be described as a weakly dominated option.

2.1.2. Follow-up questions for scenarios S1 to S7

Respondents who expressed a preference for treating either patient A or patient B in the initial question in each scenario were then asked a follow-up question. Respondents who expressed indifference between treating patient A and patient B (when such an option was available) were not asked this question.

The follow-up question was designed to identify the point at which the respondents were indifferent between treating patient A and patient B. The format of the question was similar to that used by Abel Olsen[15], and was worded as follows:

> Your choice was to treat patient [A/B], who would gain [6 months/12 months/50% health] from treatment.
> How much **shorter** would that [6 month/12 month/50% health] gain need to be for you to think that treating either patient would be **equally good**?

In each follow-up question, respondents were able to select one response from a drop-down list. If their initial choice was to treat a patient whose life would be extended by 12 months as a result of treatment, the follow-up options were: less than 1 month; 1 month; 2 months; 3 months; 4 months; 5 months; 6 months; 7 months; 8 months; 9 months; 10 months; 11 months; 12 months. If their initial choice was to treat a patient whose life would be extended by six months as a result of treatment (i.e. if they chose to treat patient A in S7), the follow-up options were: less than 1 month; 1 month; 2

months; 3 months; 4 months; 5 months; 6 months. If their initial choice was to treat a patient whose quality of life would be improved by 50% as a result of treatment, the follow-up options were: less than 10%; 10%; 20%; 30%; 40%; 50%.

In each case, the maximum value in the list of response options was equal to size of gain for the patient whose treatment the respondent had expressed preference for in the initial question. Hence, respondents were not forced to reduce the size of gain for their initially preferred patient if they did not wish to. In such cases, their response in the follow-up question could imply that they had in fact been indifferent between treating patient A and treating patient B in the initial question, even if they had been offered an indifference option (as was the case for some respondents) and had opted against choosing it.

2.1.3. Debrief questions regarding scenarios S1 to S7

Following the completion of the questions for scenarios S1 to S7, respondents were asked to indicate, using a five-point scale, the extent to which they agreed or disagreed with two statements (Likert items):

1. I found it difficult to **decide on my answers** to the questions
2. It was difficult to **understand the questions** I was asked

2.1.4. Scenario S8

Scenario S8 was included in order to examine whether an observed preference regarding an end of life premium (if any) is affected by the perspective adopted by the survey respondents (hypothesis 7). Respondents were asked to imagine that *they* could be one of the patients in need of treatment, and were presented with two possible states of the world (presented as scenario A and scenario B), each with a 50% chance of occurring. In scenario A, the respondent is 49 years old with a (just-diagnosed) life expectancy of one year (in good health) without treatment. In scenario B, the respondent is 45 years old with a (just-diagnosed) life expectancy of five years (in good health) without treatment. Scenarios A and B in S8 corresponded to the circumstances facing patient A and patient B (respectively) in S1. As with S1, a treatment taken at the time of diagnosis would generate a life extension of one year in good health, but the health service had enough funds to make the treatment available in one of the scenarios A and B, but not both.

The question posed to respondents was worded as follows:

> Suppose the health service has enough funds to make either treatment A or treatment B available, but not both. Without knowing which scenario will occur (but knowing that both have an equal chance of occurring), what would you prefer?

Respondents could respond by indicating a preference for either treatment A or treatment B being available, or by selecting an indifference option (see 2.1.6). S8 did not include a follow-up question.

The preamble for S8 acknowledged that the scenarios described may be considered unrealistic, with the intention of preventing respondents from becoming preoccupied by their hypothetical nature. This strategy is related to the use of ‘cheap talk’[16] in contingent valuation studies to mitigate the impact of hypothetical bias (where people’s stated preferences differ from their actual preferences). The purpose of cheap talk is to make respondents aware of the research question and to promote engagement, effort and attention to the choice task[17].

2.1.5. Attitudinal questions

A concern associated with stated preference studies is that it is unclear whether respondents completing abstract choice tasks would agree with the policy implications (and researchers’ interpretations) of their responses. Following the methods used by Rowen *et al.*[14]; see Rowen *et al*.[18] for full details) and Shah *et al.*[19], respondents were presented with a series of attitudinal questions intended to capture their general views about health care priority-setting, in a way that avoids the intricacies and hypothetical nature of the earlier scenario-based choice tasks.

Each attitudinal question presented a general view about priorities for the health service, and asked respondents to indicate, using a five-point scale, the extent to which they agreed or disagreed with that statement. The statements were as follows:

1. The health service should give priority to extending the life of patients who are expected to die soon as a result of a medical condition.

Agreeing with statement 1 could be interpreted as evidence of support for an end of life premium.

2. The health service should give priority to treating patients who will get the largest amount of benefit from treatment.

Agreeing with statement 2 could be interpreted as evidence of support for a QALY-maximisation approach to health care priority-setting.

3. The health service should give the same priority to treating all patients, regardless of how ill they are or when they will die.

Agreeing with statement 3 could be interpreted as evidence of a rejection of prioritisation.

4. The health service should give priority to improving the quality of life of patients who are expected to die soon as a result of a medical condition.

Agreeing with statement 4 could be interpreted as evidence of support for prioritising quality of life-improving end of life treatments over life-extending end of life treatments.

5. The health service should give priority to extending the life of patients who are expected to die soon as a result of a medical condition.

Agreeing with statement 5 could be interpreted as evidence of support for prioritising life-extending end of life treatments over quality of life-improving end of life treatments.

6. The health service should give equal priority to improving the quality of life and extending the life of patients who are expected to die soon as a result of a medical condition.

Agreeing with statement 6 could be interpreted as evidence of support for giving equal priority to quality of life-improving end of life treatments and life-extending end of life treatments. The statements were presented in two batches, with statements 1, 2 and 3 presented together first, followed by statements 4, 5 and 6 (see 2.5).

2.1.6. Study design

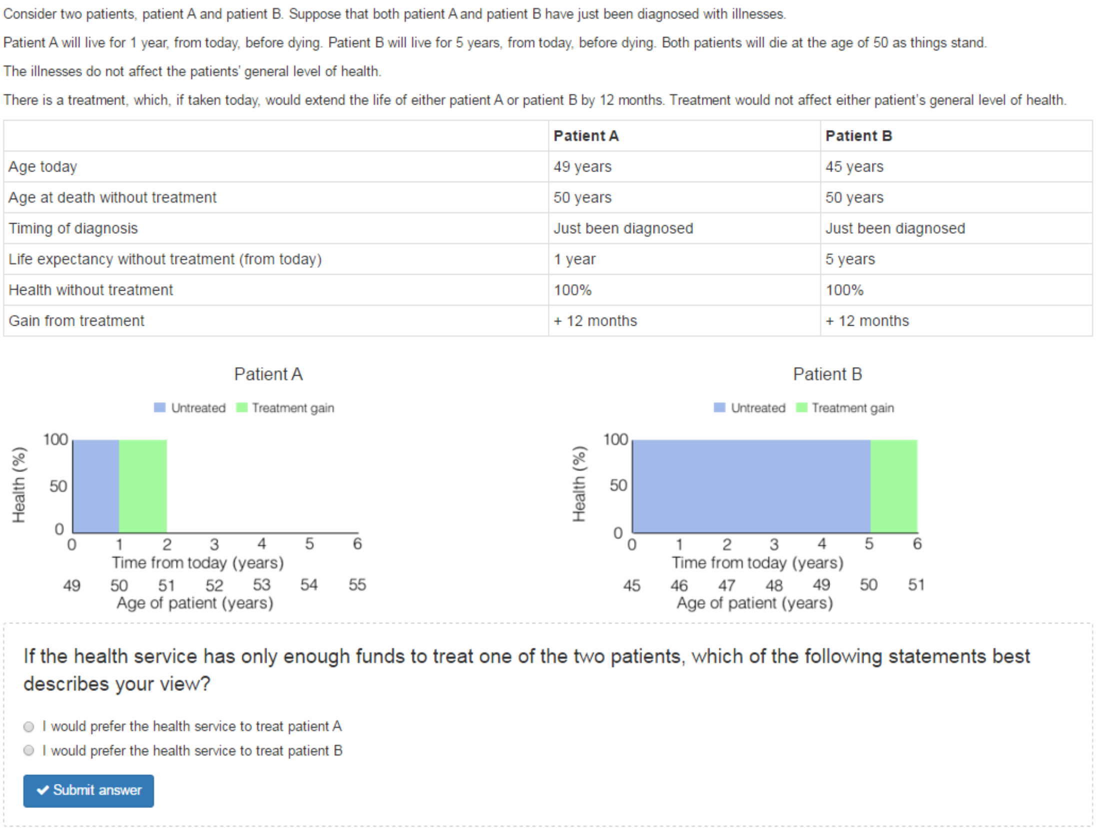
Respondents were randomly allocated to one of six versions of the survey (Table 2). In versions 4, 5 and 6, only tables and text descriptions were used to present the scenario information. In versions 1, 2 and 3, diagrams (similar to those used Shah *et al.*,[6], which in turn had adapted the design of an existing survey used by Rowen *et al.*,[14]) were used in addition to the tables and text descriptions. Visual aids were used only in the initial questions in scenarios S1 to S7; S8 and the follow-up questions in S1 to S7 did not use visual aids.

Hereafter, versions 1, 2 and 3 are referred to collectively as the ‘visual aid’ arm; and versions 4, 5 and 6 are referred to collectively as the ‘no visual aid’ arm. See Figure 1 and Figure 2 for screenshots showing how the initial question for S1 was presented in the visual aid arm and the no visual aid arm, respectively.

Table 2. Study arms and survey versions

		Visual aid arm	No visual aid arm
Forced choice arm		Version 1	Version 4
Indifference arm	Indifference option 1 arm	Version 2	Version 5
	Indifference option 2 arm	Version 3	Version 6

Figure 1. Screenshot from survey (S1; visual aid arm; forced choice arm)


Consider two patients, patient A and patient B. Suppose that both patient A and patient B have just been diagnosed with illnesses.

Patient A will live for 1 year, from today, before dying. Patient B will live for 5 years, from today, before dying. Both patients will die at the age of 50 as things stand.

The illnesses do not affect the patients' general level of health.

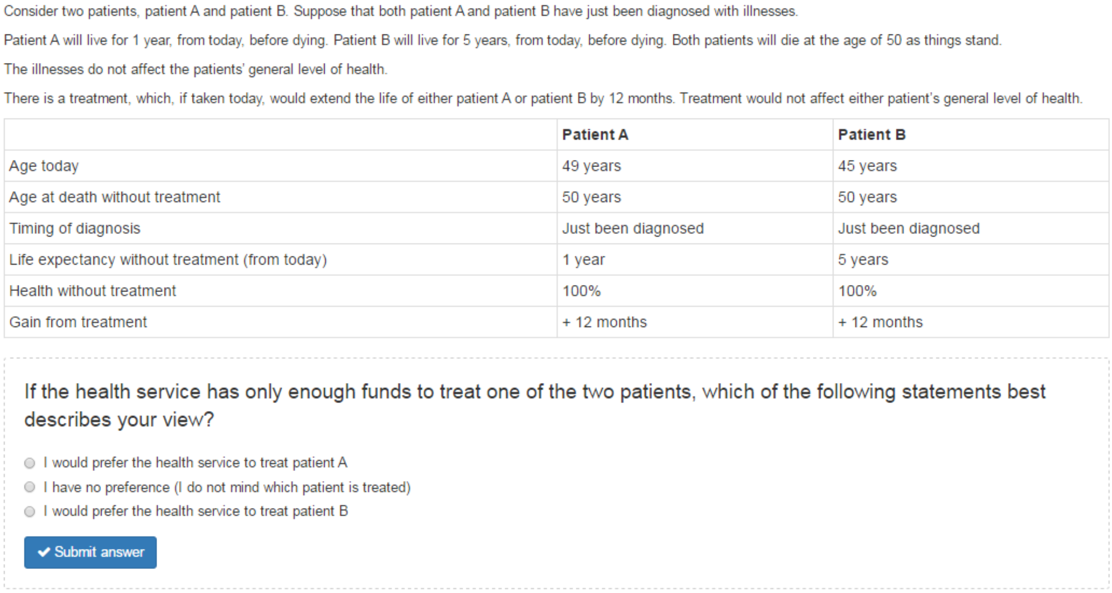
There is a treatment, which, if taken today, would extend the life of either patient A or patient B by 12 months. Treatment would not affect either patient's general level of health.

	Patient A	**Patient B**
Age today	49 years	45 years
Age at death without treatment	50 years	50 years
Timing of diagnosis	Just been diagnosed	Just been diagnosed
Life expectancy without treatment (from today)	1 year	5 years
Health without treatment	100%	100%
Gain from treatment	+ 12 months	+ 12 months

If the health service has only enough funds to treat one of the two patients, which of the following statements best describes your view?

○ I would prefer the health service to treat patient A
○ I would prefer the health service to treat patient B

Figure 2. Screenshot from survey (S1; no visual aid arm; indifference option 1 arm)


Consider two patients, patient A and patient B. Suppose that both patient A and patient B have just been diagnosed with illnesses.

Patient A will live for 1 year, from today, before dying. Patient B will live for 5 years, from today, before dying. Both patients will die at the age of 50 as things stand.

The illnesses do not affect the patients' general level of health.

There is a treatment, which, if taken today, would extend the life of either patient A or patient B by 12 months. Treatment would not affect either patient's general level of health.

	Patient A	**Patient B**
Age today	49 years	45 years
Age at death without treatment	50 years	50 years
Timing of diagnosis	Just been diagnosed	Just been diagnosed
Life expectancy without treatment (from today)	1 year	5 years
Health without treatment	100%	100%
Gain from treatment	+ 12 months	+ 12 months

If the health service has only enough funds to treat one of the two patients, which of the following statements best describes your view?

○ I would prefer the health service to treat patient A
○ I have no preference (I do not mind which patient is treated)
○ I would prefer the health service to treat patient B

In versions 1 and 4, respondents could only choose between treating patient A and treating patient B – no indifference option was available. In versions 2 and 5, an indifference option was offered, worded as follows: 'I have no preference (I do not mind which patient is treated)'. In version 3 and 6, a different indifference option was offered, worded as follows: ‘Both patients should have an equal chance of being treated (tossing a coin would be a fair way to make the choice)’. In all cases, only one of the available response options could be selected in any given scenario.

In scenario S8, all respondents, regardless of which version they had been allocated to, could choose between three options: ‘I would prefer treatment A to be available’; ‘I have no preference (I do not mind which treatment is available)’; and ‘I would prefer treatment B to be available’.

Hereafter, versions 1 and 4 are referred to collectively as the ‘forced choice’ arm; versions 2 and 5 are referred to collectively as the ‘indifference option 1’ arm; versions 3 and 6 are referred to collectively as the ‘indifference option 2’ arm; and versions 2, 3, 5 and 6 are referred to collectively as the ‘indifference’ arm. The screenshot in Figure 1 shows the choices available in the forced choice arm, and Figure 2 shows the choices available in the indifference option 1 arm.

The order in which scenarios S1 to S6 was presented was randomised for each respondent, with S7 and then S8 always following.

2.2. ADMINISTRATION OF SURVEY

An internet survey was used in favour of face-to-face interviews for this study. The main reason for this was a desire for a large sample, which is necessary in order to conduct meaningful statistical analyses and to allow respondents to be divided into multiple study arms. The budget available for data collection was insufficient for a large-sample study involving face-to-face interviews. Other benefits of internet surveys are described by Shah *et al*.[6].

2.3. SAMPLE

A target sample size of 2,400 was sought. This was determined by availability of resources and judgements that the sample needed to be sufficiently large so as to permit meaningful statistical analyses of data collected within individual arms and survey versions. The sample comprised adult members of the UK general public, who were members of a panel of a research agency, ResearchNow. Quotas and a targeted invitation strategy were used to ensure that the sample was representative of the general population in terms of selected observable characteristics: age, gender and social grade. Respondents were compensated by way of reward points which can be redeemed for gift vouchers or charity donations.

2.4. ETHICAL APPROVAL

The survey and sample recruitment procedures were reviewed and approved by the Research Ethics Committee at the School of Health and Related Research via the University of Sheffield Ethics Review Procedure.

2.5. PILOTING

A pilot was used to test a draft version of the survey in February 2016. A convenience sample of members of non-academic staff at the University of Sheffield participated in face-to-face computer-assisted personal interviews conducted by the lead author. It was made clear that the purpose of the interview was to seek feedback from respondents in order to improve the survey. In each interview, the respondent completed the draft survey on a desktop computer, with the interviewer observing but not assisting or interfering. Following an interview guide, the interviewer then asked the respondent a series of debrief questions. Each respondent was given a £10 gift voucher to thank them for their participation.

Twelve interviews were scheduled, with the intention that the six survey versions would be completed by two respondents each. Two of the respondents dropped out prior to their interviews, but it was not deemed necessary to replace them since the later interviews were not generating new insights. This meant that survey versions 1, 2, 4 and 6 were each completed by two respondents; and versions 3 and 5 were each completed by one respondent.

The pilot was completed successfully overall, with respondents mostly able to understand and complete the survey without assistance. The scenario order randomisation procedure was shown to be working as intended. Respondents required between 13 and 28 minutes to complete the survey (mean: 19.9 minutes; median: 20.0 minutes). The full interviews lasted between 23 and 39 minutes (mean: 31.7 minutes; median: 34.0 minutes). Responses to the debrief questions are summarised below.

2.5.1. Summary of responses to the debrief questions

Q1. How did you find the survey?

All but one of the 10 respondents gave positive responses to this question, stating that they found the survey enjoyable and/or interesting. The one respondent who did not enjoy the survey described it as "challenging" and referred to the "weight of responsibility" they felt whilst answering the questions.

Q2. Do you feel you understood the questions you were asked?

All of the respondents answered "yes" to this question, though two respondents added the caveat that they struggled to understand the follow-up question in each scenario. Another respondent noted that the questions required a lot of thought and that they often needed to read the text twice before they understood what was required of them.

Q3. Did the instructions adequately prepare you for the questions?

All but one of the respondents answered "yes" to this question, including the respondent who had indicated in their response to Q1 that they had not enjoyed the survey. One respondent expressed the view that the instructions did not prepare them for what was to come, but that everything made sense once they were presented with the questions themselves, and that they would not recommend making major changes to the instructions.

Q4. What did you think about the option that did not involve choosing to treat either patient A or patient B? [indifference arm only]

All of the six respondents who were asked this question indicated that the indifference option made sense to them. Three of those respondents noted that they never felt the need to choose this option, with one stating that they interpreted it as a "don't know" option.

Q5. What did you think about the diagrams used to illustrate the scenarios?

All of the five respondents who were asked this question indicated that the diagrams were clear and easy to understand, with one additionally stating that they were "really helpful". One respondent asked whether colour-blind individuals would be able to distinguish between the colours in the diagrams.

Q6. In each scenario, after choosing which patient you thought should be treated, you were asked what size that patient's gain from treatment would need to be for both patients to have equal priority. Was this clear?

Responses to this question (which 10 respondents were asked) were mixed. Four respondents said that the question was clear and easy to understand, though not necessarily easy to answer. Three respondents said that the question had confused them. Several respondents offered suggestions for improving the wording of the question. A common suggestion was to ask respondents what would make them “switch” [choices].

Q7. You were then asked to select your response from a list. Did the response you had in mind appear in this list?

Most of the respondents answered “yes” to this question. In the draft survey, the minimum response options in the follow-up questions were 1 month and 10% quality of life. Three respondents suggested including a smaller option, such as 0 months or ‘less than 1 month’, in order to capture stronger preferences.

Q8. This scenario [screenshot of S3 shown to respondent] asked you to consider one patient who has just been diagnosed with an illness and another who has known about their illness for five years. How did you find this scenario, compared to the other scenarios?

All of the respondents indicated that this scenario was easy to understand, though two noted that it was very difficult to answer. One respondent pointed out the fact that the description for one patient was longer and more detailed than that for the other, but did not suggest any changes to the text.

Q9. This scenario [screenshot of S6 shown to respondent] asked you to consider two patients whose illnesses do not affect how long they will live for, and any benefits from treatment would not take place for another 30 years. How did you find this scenario, compared to the other scenarios?

All but one of the respondents indicated that this scenario was easy to understand. Three respondents referred to the scenario as being strange and/or unrealistic. One of those respondents sought clarity about the “waves” on the graph axis (used to indicate a discontinuity in the axis).

Q10. This scenario [screenshot of S8 shown to respondent] asked you to imagine that you could be one of the patients in need of treatment, and therefore focused on your own life rather than on the lives of others. How did you find this scenario, compared to the other scenarios?

All of the respondents indicated that this scenario was easy to understand, with about half stating that it was similar to the other scenarios. One respondent described the scenario as “quite long-winded”. Another respondent suggested that the age specified in the scenarios may not be relevant to some respondents. Finally, one respondent suggested making it clearer that scenario S8 refers to the respondent’s own health whereas the preceding scenarios refer to the health of other people.

Q11. These [attitudinal] questions asked you to indicate the extent to which you agreed or disagreed with a particular statement. What did you think of these questions?

Although all of the respondents indicated that these questions were worded well and easy to understand, several noted that they had wanted to agree with all of the statements and acknowledged that they may have provided conflicting responses. Four respondents suggested displaying conflicting statements together in order to give a better sense of what might have to be given up by choosing a certain priority for the health service.

Q12. Do you have any other suggestions for improving the survey?

Two respondents suggested revising the wording of the follow-up questions in each scenario. Two respondents suggested allowing respondents to go back and revise their earlier choices. One respondent described scenarios S1 to S7 as “dehumanised”, suggesting that this could be addressed by including a cartoon image of a person to accompany the descriptions.

2.5.2. Improvements made as a result of the pilot

A number of improvements were made to the survey as a result of observations made by the interviewer and feedback provided by the pilot respondents.

Emboldening was added to highlight important words in some of the questions and text descriptions. Two feedback questions were added to the end of scenarios S1 to S7, seeking respondents’ views about how difficult they had found it to understand the questions and to decide on their answers, respectively.

The wording of the follow-up question was revised substantially. Previously, the question was worded as follows (the wording used when respondents had originally chosen to give a 12-month life extension to patient A is shown as an example):

> You indicated that you would prefer the health service to treat Patient A. We assume that if the effect of Patient A’s treatment had been

> smaller, you would have been more likely to choose to treat Patient B instead.
> What size would Patient A's gain from treatment need to be for the two patients to have equal priority?

Following the pilot, this was amended to:

> Your choice was to treat Patient A, who would gain 12 months from treatment.
> How much **shorter** would that 12 month gain need to be for you to think that treating either patient would be **equally good**?

An extra response option was added to the drop-down menus used in the follow-up questions. When respondents were choosing from a range of life extension sizes, an option of 'Less than 1' [month] was added. When respondents were choosing from a range of percentage quality of life gain sizes, an option of 'Less than 10' [%] was added.

The format of the attitudinal questions was also revised substantially. Previously, each statement was presented as a standalone question, with no opportunity to view competing (and potentially conflicting) statements. Following the pilot, the format was revised such that statements 1, 2 and 3 were presented together, and statements 4, 5 and 6 were also presented together. The statement for which a response was being sought was highlighted using emboldened text, with the other two statements greyed out but still visible.

Finally, a small number of typos were corrected.

2.6. METHODS OF ANALYSIS

Descriptive statistics were reported in order to summarise the sample, time taken to complete the survey, and responses to the scenario questions. For scenarios S1 to S7, respondents' choices were assigned to one of three categories:

A Respondent in the indifference arm indicated a preference for treating patient A; respondent in the forced choice arm indicated a preference for treating patient A and then provided a value in the follow-up question that was lower than the initial size of gain for patient A

I Respondent in the indifference arm selected the indifference option; respondent in the forced choice arm provided a value in the follow-up question that was identical to the initial size of gain for their preferred patient

B Respondent in the indifference arm indicated a preference for treating patient B; respondent in the forced choice arm indicated a preference for treating patient B and then provided a value in the follow-up question that was lower than the initial size of gain for patient B

The number and proportion of respondents in each choice category, and the number and proportion of respondents selecting each response option in the follow-up questions, was reported, both overall and by study arm.

Comparisons between arms and between scenarios were assessed using the Pearson's chi-squared test. In each case, the test was for an association between choosing to treat patient A in one scenario (or arm) and choosing to treat patient A in the other scenario (or arm). The binomial test was used to assess whether the majority of respondents chose to treat the end of life patient in S1, and whether the majority chose to provide the life-extending treatment over the quality of life-improving treatment in S4.

Two potential indicators of poor data quality or lack of respondent engagement were defined: choosing the dominated option in S7; and completing the survey in less than half of the median time taken. The impact of excluding respondents meeting one or both of these indicators was assessed, focusing on S1 (which involved choosing between treating an end of life patient and treating a non-end of life patient).

A multiple logistic regression was used to assess the impact of respondent background characteristics on the likelihood of choosing to give priority to the end of life patient in S1. The model was of the form:

$$y = X\beta + \varepsilon$$

where y is a binary dependent variable taking a value of 1 if respondents chose to treat patient A (the end of life patient) in S1, and 0 otherwise; X represents the explanatory variables; and ε represents the error term capturing other factors.

The explanatory variables included were: age (age of respondent, in whole years); gender (taking a value of 1 if respondent is male; 0 if respondent is female); social grade (taking a value of 1 if respondent is in higher social grades A, B or C1; 0 if respondent is in lower social grades C2, D or E); children (taking a value of 1 if respondent has responsibility for children; 0 if respondent does not); degree (taking a value of 1 if respondent has a degree; 0 if respondent does not); health limitations (taking a value of 0 if respondent is not limited by disability or health problems; 1 if respondent is limited ‘a little’; 2 if respondent is limited ‘a lot’); and experience of terminal illness (taking a value of 1 if respondent has had experience of terminal illness in close friends or family; 0 if respondent has not; respondents who did not wish to answer the question were coded as missing). Two binary control variables were also included to denote whether the respondent was in the forced choice arm (taking a value of 1 if respondent was in the forced choice arm; 0 if respondent was in the indifference arm) and in the visual aid arm (taking a value of 1 if respondent was in the visual aid arm; 0 if respondent was in the no visual aid arm), respectively.

A zero discount rate was assumed in all analyses. Analyses were undertaken using the Microsoft Excel and Stata[20] software packages.

3. RESULTS

Data collection was undertaken in March 2016. This commenced with a soft launch whereby the survey was closed after approximately 15% of the target number of completes had been achieved, in order to check the data for issues. No issues were observed, so the survey was re-opened until the target sample size had been achieved.

Of the 3,736 individuals who attempted to access the survey, 2,401 (64.3%) were included in the sample for analysis (Table 3). The remaining 1,335 individuals were excluded because: they did not meet the sampling quota requirements (and were therefore 'screened out'); they did not give consent to take part; they dropped out part-way through the survey; or they completed the survey in less than 271.8 seconds ('speeders'). The completion time cut-off of 271.8 seconds was one-third of the median completion time amongst the soft launch sample – it was agreed with ResearchNow that these respondents would be replaced.

Table 3. Survey completion and exclusion statistics

	n	% of all accessed	% of all screened in	% of all consents	% of all completes
Accessed	3736				
Screen-outs	89	2.4%			
Non-consents	227	6.1%	6.2%		
Non-completes	961	25.7%	26.4%	28.1%	
Speeders	58	1.6%	1.6%	1.7%	2.4%
Include in sample for analysis	2401	64.3%	65.8%	70.2%	97.6%

On average, respondents in the forced choice arm and the visual aid arm spent statistically significantly longer on the survey than did respondents in the indifference arm and the no visual aid arm, respectively (Table 4) (forced choice vs. indifference: Welch's t-test; p<0.01; visual aid vs. no visual aid: Welch's t-test; p<0.01).

Table 4. Time taken (in minutes) to complete survey

	Overall (n=2,401)	Forced choice (n=807)	Indifference (n=1,594)	Visual aid (n=1,202)	No visual aid (n=1,199)
Mean	16.7	17.7	16.3	17.6	15.9
Median	14.1	15.0	13.8	14.9	13.5
SD	10.9	11.6	10.5	11.4	10.2

Table 5 presents the background characteristics of the sample. The sample was representative of the general population with respect to age, gender and social grade[21]. The study arms were well balanced in terms of their composition.

Table 5. Sample background characteristics

		Sample (all versions)		Forced choice (ver. 1,4)		Indifference (ver. 2,3,5,6)		Visual aid (ver. 1,2,3)		No visual aid (ver. 4,5,6)		Population
		n	%	n	%	n	%	n	%	n	%	%
Total		2401	100.0%	807	100.0%	1594	100.0%	1202	100.0%	1199	100.0%	
Age (years)	18-29	477	19.9%	165	20.4%	312	19.6%	241	20.0%	236	19.7%	21%
	30-44	633	26.4%	225	27.9%	408	25.6%	323	26.9%	310	25.9%	26%
	45-59	597	24.9%	178	22.1%	419	26.3%	302	25.1%	295	24.6%	25%
	60+	694	28.9%	239	29.6%	455	28.5%	336	28.0%	358	29.9%	28%
Gender	Female	1235	51.4%	429	53.2%	806	50.6%	615	51.2%	620	51.7%	51%
	Male	1166	48.6%	378	46.8%	788	49.4%	587	48.8%	579	48.3%	49%
Social grade [a]	A	93	3.9%	26	3.2%	67	4.2%	42	3.5%	51	4.3%	4%
	B	534	22.2%	176	21.8%	358	22.5%	271	22.5%	263	21.9%	23%
	C1	745	31.0%	260	32.2%	485	30.4%	381	31.7%	364	30.4%	27%
	C2	525	21.9%	174	21.6%	351	22.0%	267	22.2%	258	21.5%	21%
	D	290	12.1%	97	12.0%	193	12.1%	138	11.5%	152	12.7%	16%
	E	214	8.9%	74	9.2%	140	8.8%	103	8.6%	111	9.3%	9%
Household composition	With children	765	31.9%	245	30.4%	520	32.6%	388	32.3%	377	31.4%	
	Without children	1636	68.1%	562	69.6%	1074	67.4%	814	67.7%	822	68.6%	
Education	None beyond min. school leaving age	559	23.3%	181	22.4%	378	23.7%	286	23.8%	273	22.8%	
	Beyond min. school leaving age; no degree	768	32.0%	243	30.1%	525	32.9%	387	32.2%	381	31.8%	
	Beyond min. school leaving age; degree	1074	44.7%	383	47.5%	691	43.4%	529	44.0%	545	45.5%	
Self-reported general health	Very good	507	21.1%	181	22.4%	326	20.5%	248	20.6%	259	21.6%	
	Good	1144	47.6%	377	46.7%	767	48.1%	588	48.9%	556	46.4%	
	Fair	575	23.9%	192	23.8%	383	24.0%	291	24.2%	284	23.7%	
	Poor	157	6.5%	52	6.4%	105	6.6%	71	5.9%	86	7.2%	
	Very poor	18	0.7%	5	0.6%	13	0.8%	4	0.3%	14	1.2%	
Experience of terminal illness in friends/family	Yes	1513	63.0%	507	62.8%	1006	63.1%	766	63.7%	747	62.3%	
	No	803	33.4%	277	34.3%	526	33.0%	394	32.8%	409	34.1%	
	Question skipped	85	3.5%	23	2.9%	62	3.9%	42	3.5%	43	3.6%	

[a] Refers to the occupation/qualifications/responsibilities of the chief wage earner of the respondent's household; see National Readership Survey[22].

3.1. AGGREGATE RESPONSES TO SCENARIO QUESTIONS

Table 6 to Table 13 report the aggregate response data for each scenario, both overall and by study arm. In each column in these tables, the modal choice is emboldened.

Table 6 shows that in S1, the most common choice overall was to express indifference. The proportion of respondents choosing to treat the end of life patient is statistically significantly different from (less than) 50% (binomial test; one-sided test: p<0.01; two-sided test: p<0.01). Hence, the hypothesis that people place no more weight on a unit of health gain for end of life patients as on that for other types of patients (hypothesis 1) *cannot be rejected*.

Table 6. S1: End of life patient (A) vs. non-end of life patient (B)

	Overall	Forced choice	Indifference option 1	Indifference option 2	No visual aid	Visual aid
A	765 (31.9%)	**316 (39.2%)**	238 (29.8%)	211 (26.5%)	380 (31.7%)	385 (32.0%)
I	**833 (34.7%)**	206 (25.5%)	**298 (37.3%)**	**329 (41.3%)**	**440 (36.7%)**	393 (32.7%)
B	803 (33.4%)	285 (35.3%)	262 (32.8%)	256 (32.2%)	379 (31.6%)	**424 (35.3%)**
Total	2401 (100.0%)	807 (100.0%)	798 (100.0%)	796 (100.0%)	1199 (100.0%)	1202 (100.0%)

Table 7 shows that treating a 45 year old patient with five years of life expectancy without treatment was preferred by the majority of respondents to treating a 69 year old patient with one year of life expectancy without treatment. The proportion of respondents choosing the former option represents the largest majority across all scenarios. Scenario S2 is one of only two scenarios (the other being S5) with a robust modal response across all study arms.

Table 7. S2: Older end of life patient (A) vs. younger non-end of life patient (B)

	Overall	Forced choice	Indifference option 1	Indifference option 2	No visual aid	Visual aid
A	340 (14.2%)	140 (17.3%)	107 (13.4%)	93 (11.7%)	168 (14.0%)	172 (14.3%)
I	623 (25.9%)	213 (26.4%)	187 (23.4%)	223 (28.0%)	316 (26.4%)	307 (25.5%)
B	**1438 (59.9%)**	**454 (56.3%)**	**504 (63.2%)**	**480 (60.3%)**	**715 (59.6%)**	**723 (60.1%)**
Total	2401 (100.0%)	807 (100.0%)	798 (100.0%)	796 (100.0%)	1199 (100.0%)	1202 (100.0%)

Comparing Table 6 with Table 8 shows that when the end of life patient was revealed to have known about their prognosis for some time (as in S3), there was a slight shift towards preferring to treat the non-end of life patient who has only just learned of their prognosis. As in S1, however, the most common choice in S3 overall was to express indifference.

Table 8. S3: End of life patient with more time with knowledge (A) vs. non-end of life patient with less time with knowledge (B)

	Overall	Forced choice	Indifference option 1	Indifference option 2	No visual aid	Visual aid
A	579 (24.1%)	254 (31.5%)	169 (21.2%)	156 (19.6%)	277 (23.1%)	302 (25.1%)
I	**914 (38.1%)**	191 (23.7%)	**344 (43.1%)**	**379 (47.6%)**	458 (38.2%)	**456 (37.9%)**
B	908 (37.8%)	**362 (44.9%)**	285 (35.7%)	261 (32.8%)	**464 (38.7%)**	444 (36.9%)
Total	2401 (100.0%)	807 (100.0%)	798 (100.0%)	796 (100.0%)	1199 (100.0%)	1202 (100.0%)

Table 9 shows that more respondents chose the quality of life-improving treatment than the life-extending treatment in S4, though this preference was less pronounced amongst respondents in the visual aid arm. Overall, the proportion of respondents choosing to provide the life-extending treatment is statistically significantly different from (less than) 50% (binomial test; one-sided test: p<0.01; two-sided test: p<0.01). Hence, the hypothesis that people place no more weight on life-extending treatments than on quality of life-improving treatments for end of life patients (hypothesis 4) *cannot be rejected*.

Table 9. S4: Quality of life improvement for end of life patient (A) vs. life extension for end of life patient (B)

	Overall	Forced choice	Indifference option 1	Indifference option 2	No visual aid	Visual aid
A	**969 (40.4%)**	**350 (43.4%)**	**325 (40.7%)**	294 (36.9%)	**522 (43.5%)**	**447 (37.2%)**
I	817 (34.0%)	196 (24.3%)	288 (36.1%)	**333 (41.8%)**	413 (34.4%)	404 (33.6%)
B	615 (25.6%)	261 (32.3%)	185 (23.2%)	169 (21.2%)	264 (22.0%)	351 (29.2%)
Total	2401 (100.0%)	807 (100.0%)	798 (100.0%)	796 (100.0%)	1199 (100.0%)	1202 (100.0%)

Comparing Table 9 with Table 10 shows that when the life-extending treatment was for a non-end of life patient (as in S5) rather than for an end of life patient (as in S4), respondents were more likely to choose the life-extending treatment and less likely to express indifference.

Table 10. S5: Quality of life improvement for end of life patient (A) vs. life extension for non-end of life patient (B)

	Overall	Forced choice	Indifference option 1	Indifference option 2	No visual aid	Visual aid
A	**924 (38.5%)**	**330 (40.9%)**	**300 (37.6%)**	**294 (36.9%)**	**464 (38.7%)**	**460 (38.3%)**
I	707 (29.4%)	191 (23.7%)	248 (31.1%)	268 (33.7%)	374 (31.2%)	333 (27.7%)

B	770 (32.1%)	286 (35.4%)	250 (31.3%)	234 (29.4%)	361 (30.1%)	409 (34.0%)
Total	2401 (100.0%)	807 (100.0%)	798 (100.0%)	796 (100.0%)	1199 (100.0%)	1202 (100.0%)

Table 11 shows that that the preference for quality of life-improving treatments over life-extending treatments exists not only in the end of life context (as in S4) but also in a non-end of life context where the benefits from treatment would occur at the end of the patient's normal life expectancy (as in S6). In the indifference arm, however, the most common choice was to express indifference.

Table 11. S6: Quality of life improvement at end of normal life expectancy (A) vs. life extension at end of normal life expectancy (B)

	Overall	Forced choice	Indifference option 1	Indifference option 2	No visual aid	Visual aid
A	**1024 (42.6%)**	**401 (49.7%)**	323 (40.5%)	300 (37.7%)	**534 (44.5%)**	**490 (40.8%)**
I	891 (37.1%)	203 (25.2%)	**348 (43.6%)**	**340 (42.7%)**	466 (38.9%)	425 (35.4%)
B	486 (20.2%)	203 (25.2%)	127 (15.9%)	156 (19.6%)	199 (16.6%)	287 (23.9%)
Total	2401 (100.0%)	807 (100.0%)	798 (100.0%)	796 (100.0%)	1199 (100.0%)	1202 (100.0%)

Table 12 shows that the vast majority of respondents did not choose the dominated (smaller life extension) option in S7, though many expressed indifference – particularly in the indifference arm.

Table 12. S7: Smaller life extension for end of life patient (A) vs. larger life extension for end of life patient (B)

	Overall	Forced choice	Indifference option 1	Indifference option 2	No visual aid	Visual aid
A	190 (7.9%)	82 (10.2%)	62 (7.8%)	46 (5.8%)	104 (8.7%)	86 (7.2%)
I	866 (36.1%)	165 (20.4%)	321 (40.2%)	**380 (47.7%)**	451 (37.6%)	415 (34.5%)
B	**1345 (56.0%)**	**560 (69.4%)**	**415 (52.0%)**	370 (46.5%)	**644 (53.7%)**	**701 (58.3%)**
Total	2401 (100.0%)	807 (100.0%)	798 (100.0%)	796 (100.0%)	1199 (100.0%)	1202 (100.0%)

Comparing Table 6 and Table 13 shows that a larger proportion of respondents expressed preference for the provision of treatment for the non-end of life patient when answering from an individual perspective (as in S8) rather than from a social decision-maker perspective (as in S1). It should be noted that visual aids were not used in S8 (for any respondent), and an indifference option was always available (for all respondents). Differences in choices across arms were minimal in comparison to some of the other scenarios.

Table 13. S8: Individual perspective adaptation of S1 – 1 year of life expectancy without treatment (A) vs. 5 years of life expectancy without treatment (B)

	Overall	Forced choice	Indifference option 1	Indifference option 2	No visual aid	Visual aid
A	440 (18.3%)	172 (21.3%)	134 (16.8%)	134 (16.8%)	212 (17.7%)	228 (19.0%)
I	970 (40.4%)	**326 (40.4%)**	320 (40.1%)	324 (40.7%)	492 (41.0%)	478 (39.8%)
B	**991 (41.3%)**	309 (38.3%)	**344 (43.1%)**	**338 (42.5%)**	**495 (41.3%)**	**496 (41.3%)**
Total	2401 (100.0%)	807 (100.0%)	798 (100.0%)	796 (100.0%)	1199 (100.0%)	1202 (100.0%)

3.2. IMPACT OF EXPERIMENTAL MODIFICATIONS

3.2.1. Impact of including an indifference option

Across scenarios S1 to S8, indifference was expressed 26.2% of the time by respondents in the forced choice arm; and 38.7% of the time by respondents in the indifference arm. Respondents in the indifference arm were statistically significantly more likely than respondents in the forced choice arm to express indifference (chi-squared test; p<0.01). This tendency is observed in all scenarios except S2 and S8, in which the proportions of respondents expressing indifference did not differ greatly by arm.

Further, in S1 the modal choice was to treat the end of life patient amongst respondents in the forced choice arm, whereas for respondents in the indifference arm this was the least common choice. The association between the availability of an indifference option and the propensity to choose to treat the end of life patient is statistically significant (chi-squared test; p<0.01). Hence, the hypothesis that preferences regarding an end of life premium are unaffected by whether an indifference option is included (hypothesis 9) *is rejected*.

The propensity to express indifference did not only differ between the indifference arm and the force choice arm, but also between the two indifference option arms. Across scenarios S1 to S7 (S8 is disregarded as the wording of the indifference option was the same for all respondents in that scenario), the indifference option was chosen 36.4% of the time by respondents in indifference option 1 arm; and 40.4% of the time by respondents in the indifference option 2 arm. Respondents in the indifference option 2 arm were statistically significantly more likely than respondents in the indifference option 1 arm to express indifference (chi-squared test; p<0.01). This tendency is observed in all scenarios except S6, in which the proportions of respondents expressing indifference did not differ greatly (or statistically significantly) by arm.

3.2.2. Impact of including a visual aid

The impact of including a visual aid varied across scenarios. In each of the three scenarios in which one of the options involved a quality of life-improving treatment (S4, S5, S6), respondents in the visual aid arm were more likely than respondents in the no visual aid arm to choose the life-extending treatment over the quality of life-improving treatment. In two of those scenarios, the association between study arm and response pattern was statistically significant at the 5% level (chi-squared test; S4: $p<0.01$; S5: $p=0.07$; S6: $p<0.01$).

In the other four scenarios, the patterns of responses did not differ greatly between arms. The association between study arm and response pattern was not statistically significant at the 5% level in these scenarios (chi-squared test; S1: $p=0.07$; S2: $p=0.90$; S3: $p=0.47$; S7: $p=0.06$). Whilst the hypothesis that preferences regarding an end of life premium are unaffected by whether visual aids are included (hypothesis 8) *cannot be rejected* on the basis of S1 alone, it is clear that the results of stated priority-setting preference studies are to some extent influenced by whether the information is presented diagrammatically or not.

3.3. RESPONSES TO FOLLOW-UP QUESTIONS

In scenarios S1 to S6, the most common follow-up responses made by respondents who initially expressed a preference for a 12-month life extension were:

- To choose a gain half the size of the initial gain – i.e. 6-month life extension
- To choose a gain equal in size to the initial gain – i.e. 12-month life extension
- To choose the smallest gain possible – i.e. life extension of 1 month or less

Note that respondents in the indifference arm answered the follow-up question only if they had initially chosen to treat either patient A or patient B (not if they had chosen the indifference option). Yet a sizeable minority of those respondents returned the same size of gain in the follow-up question, indicating either that they were indifferent between the two (but for whatever reason had not wanted to choose the initial indifference option) or that the follow-up response options were too crude (e.g. they might have selected 11.5 months). A caveat here is that it was not made explicit to respondents that one of the response options in the follow-up question was equivalent to choosing the indifference option in the initial question. Nevertheless, it is notable that the proportion of respondents returning the same size of gain was generally not much smaller, and in some cases greater, in the indifference arm compared to the forced choice arm.

In scenarios S4 to S6, no ‘mid-point’ value (i.e. 25% gain) was available for respondents who initially chose the quality of life-improving treatment over the life-extending treatment. In these scenarios, a more even spread of responses across the available options was observed.

In scenario S7, the most common follow-up response, by some distance, was to choose a 6-month life-extension. With the exception of S7, the pattern of responses was very similar across scenarios (Table 14) and across arms.

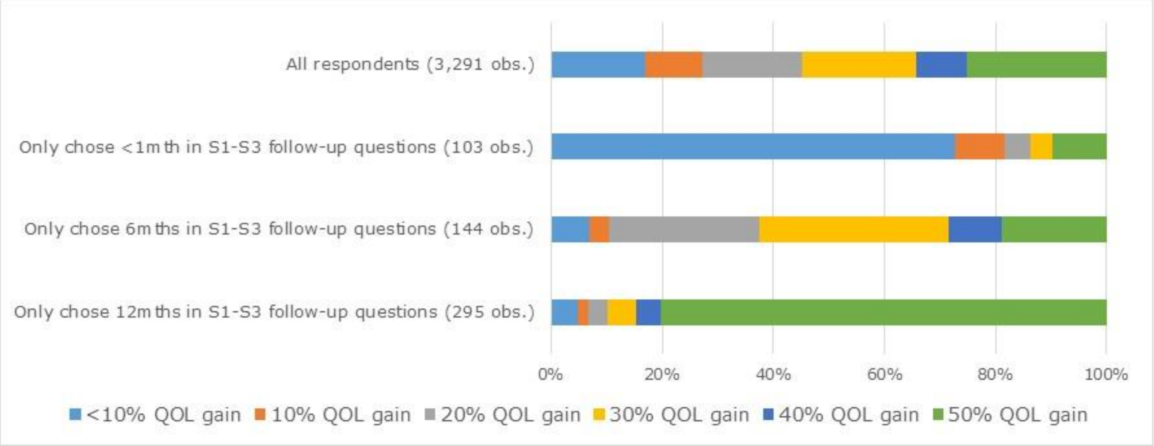
Table 14. Summary of responses to follow-up questions, by scenario

Scenario	Initial choice		Follow-up choice - size of gain		
	Choice	Gain	Mean	Median	Mode
S1	A	12 months	5.9	6.0	6.0
	B	12 months	6.8	6.0	12.0
S2	A	12 months	6.3	6.0	12.0
	B	12 months	6.4	6.0	12.0
S3	A	12 months	6.2	6.0	6.0
	B	12 months	6.6	6.0	12.0
S4	A	50%	27.4	30.0	50.0
	B	12 months	5.8	6.0	6.0
S5	A	50%	26.7	30.0	50.0
	B	12 months	6.0	6.0	6.0
S6	A	50%	26.9	30.0	50.0
	B	12 months	6.2	6.0	6.0
S7	A	6 months	3.8	4.0	6.0
	B	12 months	6.3	6.0	6.0

The way in which respondents responded to the follow-up questions in scenarios S1 to S3 (which, regardless of the initial choice, always involved selecting a life extension of 12 months or shorter) is associated with the way in which they responded to the follow-up questions in scenarios S4 to S6. Figure 3shows the distribution of responses to the follow-up questions in S4 to S6, for respondents who initially chose the quality of life improvement option in those scenarios. The follow-up questions for these respondents involved selecting a quality of life improvement of 50% or smaller, with no obvious mid-point available. The upper bar represents the data for all respondents who chose the quality of life improvement option at least once in S4, S5 and S6, regardless of their responses in the other scenarios (3,291 observations). No tendency towards one particular follow-up response option is observed.

The lower three bars show the same data for subgroups of respondents who appear to have followed some heuristic in their responses to scenarios S1 to S3. The majority of respondents who selected the minimum possible life extension (‘less than one month’) in S1, S2 and S3 also selected the minimum possible quality of life improvement (‘less than 10%’) in S4, S5 and S6. The majority of respondents who selected the maximum possible life extension (12 months) in S1, S2 and S3 also selected the maximum possible quality of life improvement (50%) in S4, S5 and S6. The responses of respondents who selected the mid-point life extension (6 months) in S1, S2 and S3 were more evenly spread, though the options closest to the mid-point of 25% (20% and 30%) were selected most frequently.

Figure 3. Responses to S4-S6 follow-up questions made by respondents who initially chose the quality of life improvement option in S4-S6



3.4. SETS OF CHOICES

There are 2,187 (=3^7) different combinations of choices that respondents could have made in scenarios S1 to S7, of which 784 were made by at least one respondent in the sample. The five most common sets of choices (covering 16.5% of respondents) are presented in Table 15, together with possible (face-value) explanations of those choices.

Table 15. Five most common sets of choices in S1 to S7

Set of choices [a]	Count	Possible explanations
III-III-I	189	Rejection of prioritisation based on attributes presented Not enough difference between patients / options to justify prioritising one Lack of engagement / shortcutting the survey
BBB-AAA-B	56	Rejection of end of life premium; priority to quality of life improvement over life extension (in end of life and non-end of life contexts); QALY-max when all other attributes levels are equal
BBB-ABA-B	55	Rejection of end of life premium; priority to quality of life improvement over life extension (in end of life and non-end of life contexts) but preference for treating non-end of life patient outweighs preference for quality of life improvement; QALY-max when all other attributes levels are equal
ABB-AAA-B	50	Support for end of life premium; priority to younger patients and patients with less time to prepare (both of which outweigh the preference for end of life); priority to quality of life improvement over life extension (in end of life and non-end of life contexts); QALY-max when all other attributes levels are equal
ABA-AAA-B	46	Support for end of life premium; priority to younger patients (outweighing the preference for end of life); priority to quality of life improvement over life extension (in end of life and non-end of life contexts); QALY-max when all other attributes levels are equal

[a] For ease of readability, each set of choices has been presented so that S1, S2 and S3 (in which treatment would generate life extensions for both patients) are grouped together; and S4, S5 and S6 (in which treating patient A would generate a quality of life improvement) are also grouped together.

Choices that would be most consistent with NICE’s end of life policy are as follows:

- S1: A (priority to end of life patient)
- S2: A (priority to end of life patient, regardless of age)
- S3: A (priority to end of life patient, regardless of time of diagnosis)
- S4: B (priority to life-extending treatment for end of life patient)
- S5: N/A (neither option would meet the NICE criteria)
- S6: N/A (neither option would meet the NICE criteria)
- S7: B (QALY-maximisation)

Thirty-one of the 2,401 respondents (1.3%) responded to scenarios S1, S2, S3, S4 and S7 in such a way that would be entirely consistent with NICE’s end of life policy.

An alternative (and less prescriptive) end of life policy, which involves always giving priority to the end of life patient but does not impose any restrictions about the size or type (i.e. quality of life improvement or life extension) of gain, would be consistent with the following choices:

- S1: A (priority to end of life patient)
- S2: A (priority to end of life patient, regardless of age)
- S3: A (priority to end of life patient, regardless of time of diagnosis)
- S4: N/A (both patients are equally at the end of life)
- S5: A (priority to end of life patient, regardless of type of gain)
- S6: N/A (both patients are non-end of life)
- S7: N/A (both patients are equally at the end of life)

Seventy-one of the 2,401 respondents (3.0%) responded to scenarios S1, S2, S3 and S5 in such a way that would be entirely consistent with this alternative end of life policy.

A pure QALY-maximiser (with zero time preference) who is not concerned about the recipient of the QALYs should make the following set of choices: III-III-B. Sixteen of the 2,401 respondents (0.7%) made this set of choices. Table 16 shows how respondents who expressed indifference in all seven scenarios (III-III-I) differed from those interpreted as pure QALY-maximisers (III-III-B) in terms of selected statistics.

Table 16. Selected statistics for respondents expressing indifference in all scenarios (III-III-I) and respondents who choices reflect QALY-maximisation (III-III-B)

		III-III-I	III-III-B
Number of respondents	n	189	16
Arm	No visual aid	97 (51.3%)	8 (50.0%)
	Visual aid	92 (48.7%)	8 (50.0%)
Time taken to complete survey	Median (min)	9.3	22.1
	<423 sec [a]	41 (21.7%)	1 (6.3%)
	>423 sec	148 (78.3%)	15 (93.8%)
Agreement with attitudinal statement: "The health service should give priority to treating patients who will get the largest amount of benefit from treatment"	Strongly or moderately agree [b]	73 (38.6%)	12 (75.0%)

[a] 423 seconds is half of the median time taken by all respondents in the sample. See 3.7 for an exploration of the use of this statistic as an indicator of data quality.
[b] 72.4% of all respondents in the sample agreed (either strongly or moderately) with this statement – see 3.6 for analysis.

The number of respondents in the III-III-B group is very small, which limits any conclusions that can be drawn from this analysis. Nevertheless, both groups are about equally split between the visual aid and no visual aid arms. Respondents in the III-III-I group spent much less time completing the survey than those in the III-III-B group (and also less than the average respondent). The majority of respondents in the III-III-B group expressed agreement with the attitudinal statement that most closely reflects the QALY-maximisation stance, whereas respondents in the III-III-I were much less likely *than average* to agree with this statement.

3.5. CROSS-TABULATIONS OF RESPONSES FROM SELECTED PAIRS OF SCENARIOS

Table 17 to Table 22 provide cross-tabulations of response data from selected combinations of scenarios that can be used to test some of the hypotheses set out 6.1. In these tables, the shaded cells refer to respondents whose choice (A, I or B) was the same in both scenarios, and the sums of the shaded cells are reported as table footnotes.

Table 17. Cross-tabulation – S1 vs. S2

		S2			
		A	I	B	Total
S1	A	198 (8.2%)	75 (3.1%)	492 (20.5%)	765 (31.9%)
	I	56 (2.3%)	435 (18.1%)	342 (14.2%)	833 (34.7%)
	B	86 (3.6%)	113 (4.7%)	604 (25.2%)	803 (33.4%)
	Total	340 (14.2%)	623 (25.9%)	1438 (59.9%)	2401 (100.0%)

Sum of shaded cells: 51.5%

The association between patient age and the propensity to prioritise the treatment of the end of life patient is statistically significant (chi-squared test; p<0.01). Hence, the hypothesis that preferences regarding an end of life premium are unaffected by whether or not the end of life patient is older than the non-end of life patient (hypothesis 2) *is rejected*.

Table 18. Cross-tabulation – S1 vs. S3

		S3			
		A	I	B	Total
S1	A	367 (15.3%)	150 (6.2%)	248 (10.3%)	765 (31.9%)
	I	92 (3.8%)	577 (24.0%)	164 (6.8%)	833 (34.7%)
	B	120 (5.0%)	187 (7.8%)	496 (20.7%)	803 (33.4%)
	Total	579 (24.1%)	914 (38.1%)	908 (37.8%)	2401 (100.0%)

Sum of shaded cells: 60.0%

Of the 2,401 respondents in the sample, 1,440 (60.0%) made the same choices – i.e. AA, II or BB – in both S1 and S3. The association between time with knowledge and the propensity to prioritise the treatment of the end of life patient is statistically significant (chi-squared test; p<0.01). Hence, the hypothesis that preferences regarding an end of life premium are unaffected by whether or not the end of life patient has known about their prognosis for longer than the non-end of life patient (hypothesis 3) *is rejected*.

Table 19. Cross-tabulation – S1 vs. S5

		S5			
		A	I	B	Total
S1	A	446 (18.6%)	113 (4.7%)	206 (8.6%)	765 (31.9%)
	I	209 (8.7%)	465 (19.4%)	159 (6.6%)	833 (34.7%)
	B	269 (11.2%)	129 (5.4%)	405 (16.9%)	803 (33.4%)
	Total	924 (38.5%)	707 (29.4%)	770 (32.1%)	2401 (100.0%)

Sum of shaded cells: 54.9%

The association between type of end of life treatment (quality of life improvement or life extension) and the propensity to prioritise the treatment of the end of life patient is statistically significant (chi-squared test; p<0.01). Hence, the hypothesis that preferences regarding an end of life premium are unaffected by whether the end of life treatment is quality of life-improving or life-extending (hypothesis 5) *is rejected*.

Table 20. Cross-tabulation – S4 vs. S6

		S6			
		A	I	B	Total
S4	A	601 (25.0%)	218 (9.1%)	150 (6.2%)	969 (40.4%)
	I	188 (7.8%)	540 (22.5%)	89 (3.7%)	817 (34.0%)
	B	235 (9.8%)	133 (5.5%)	247 (10.3%)	615 (25.6%)
	Total	1024 (42.6%)	891 (37.1%)	486 (20.2%)	2401 (100.0%)

Sum of shaded cells: 57.8%

The association between context (end of life or non-end of life) and the propensity to prioritise the quality of life-improving treatment is not statistically significant (chi-squared test; p=0.11). Hence, the hypothesis that preferences between quality of life improvements and life extensions are unaffected by whether the gain occurs in an end of life or non-end of life context (hypothesis 6) *cannot be rejected*.

Table 21. Cross-tabulation – S1 vs. S8

		S8			
		A	I	B	Total
S1	A	280 (11.7%)	257 (10.7%)	228 (9.5%)	765 (31.9%)
	I	97 (4.0%)	460 (19.2%)	276 (11.5%)	833 (34.7%)
	B	63 (2.6%)	253 (10.5%)	487 (20.3%)	803 (33.4%)
	Total	440 (18.3%)	970 (40.4%)	991 (41.3%)	2401 (100.0%)

Sum of shaded cells: 51.2%

The association between study perspective (individual or social decision-maker) and the propensity to prioritise provision of the end of life treatment is statistically significant (chi-squared test; p<0.01). Hence, the hypothesis that preferences regarding an end of life premium are unaffected by whether the preferences are being elicited from an individual or a social decision perspective (hypothesis 7) *is rejected*.

Note that scenario S8 differed from the other scenarios in that there was no visual aid (even for respondents in the visual aid arm), and indifference option 1 was used (even for respondents in the forced choice or indifference option 2 arms). To control for the effects of these design choices, the above analysis is repeated only for respondents who completed survey version 5 (no visual aid; indifference option 1), for whom the difference in framing between S8 and the other scenarios was least pronounced.

Table 22. Cross-tabulation – S1 vs. S8 (survey version 5 only)

		S8			
		A	I	B	Total
S1	A	42 (10.6%)	40 (10.1%)	47 (11.8%)	129 (32.5%)
	I	16 (4.0%)	101 (25.4%)	41 (10.3%)	158 (39.8%)
	B	9 (2.3%)	24 (6.0%)	77 (19.4%)	110 (27.7%)
	Total	67 (16.9%)	165 (41.6%)	165 (41.6%)	397 (100.0%)

Sum of shaded cells: 55.4%

As above, the association between study perspective and the propensity to prioritise provision of the end of life treatment is statistically significant (chi-squared test; $p<0.01$).

A further null hypothesis of relevance when comparing S1 and S8 is that the propensity to *express indifference* is unaffected by the perspective adopted. The alternative hypothesis is that when making choices from an individual perspective, respondents are more likely to express indifference, possibly in an attempt to minimise regret – i.e. disutility from learning that they would have been better off having not taken the action they did.

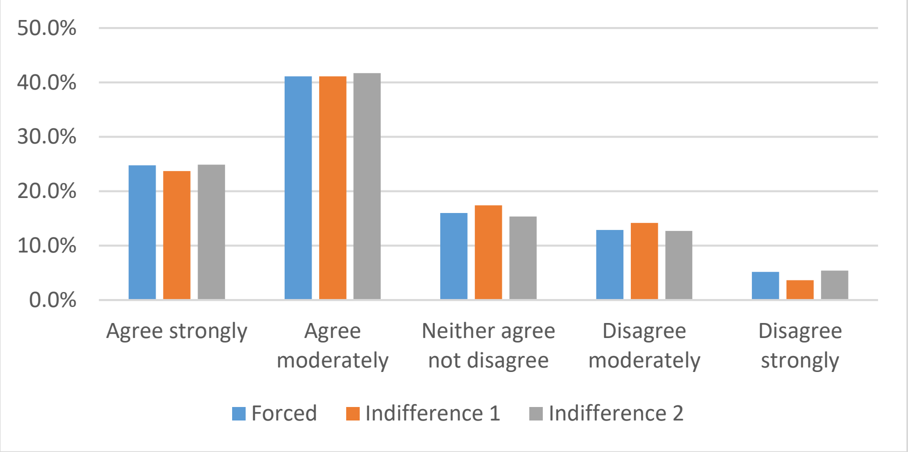
Although a larger proportion of survey version 5 respondents expressed indifference in S8 (165 respondents; 41.6%) than in S1 (158 respondents; 39.8%), the association between perspective and the propensity to express indifference is not statistically significant (chi-squared test; $p=0.61$). Hence, the hypothesis that the propensity to express indifference is unaffected by the perspective adopted *cannot be rejected*.

3.6. DEBRIEF STATEMENTS

Overall, the majority of respondents agreed with debrief statement 1 (65.8% agreed either strongly or moderately with this statement) and disagreed with debrief statement 2 (57.4% disagreed either strongly or moderately with this statement).

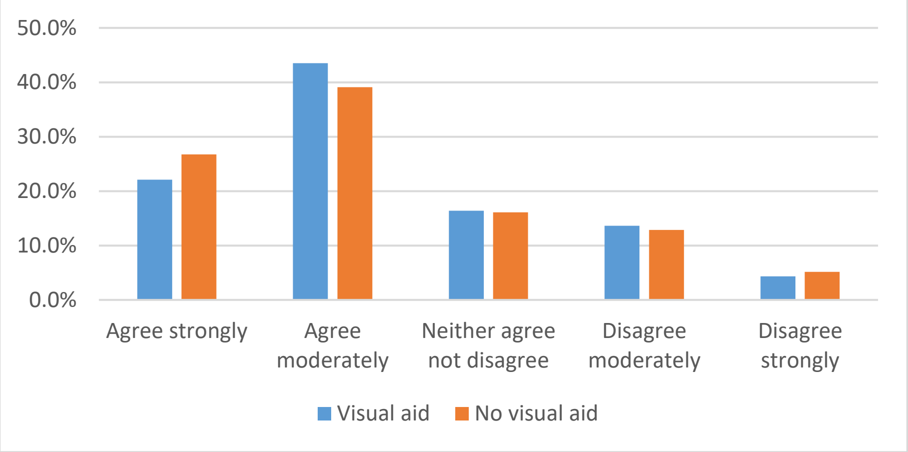
The extent to which respondents agreed or disagreed with debrief statement 1 did not vary greatly by study arm (Figure 4 and Figure 5). On the other hand, respondents in the forced choice arm were more likely to agree with debrief statement 2 than those who were given an indifference option (Figure 6).

Figure 4. Responses to debrief statement 1 (by indifference arm)



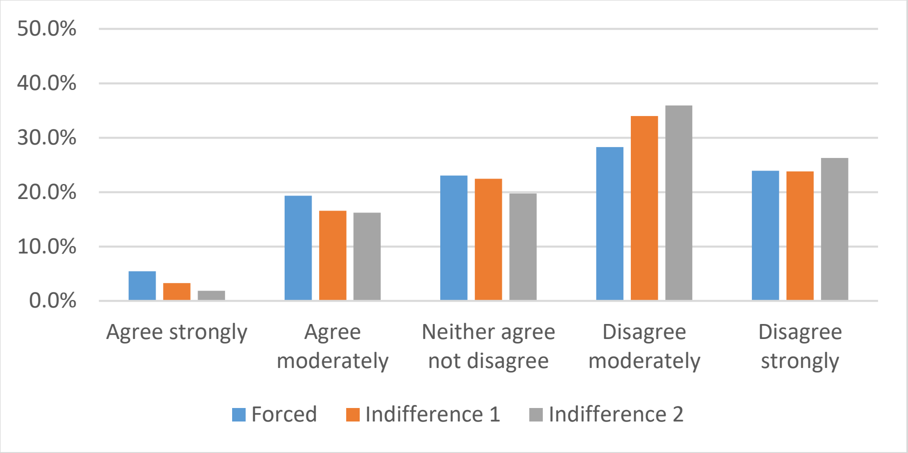
Statement 1: I found it difficult to decide on my answers to the questions

Figure 5. Responses to debrief statement 1 (by visual aid arm)



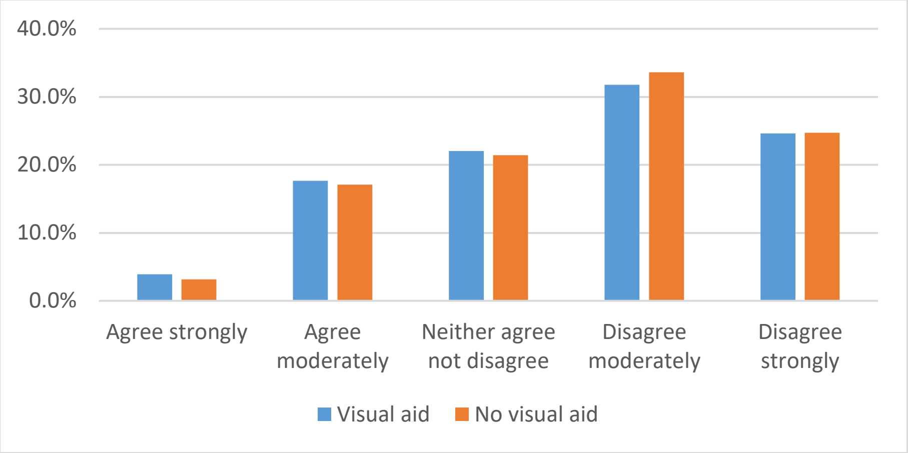
Statement 1: I found it difficult to decide on my answers to the questions

Figure 6. Responses to debrief statement 2 (by indifference arm)



Statement 2: It was difficult to understand the questions I was asked

Figure 7. Responses to debrief statement 2 (by visual aid arm)



Statement 2: It was difficult to understand the questions I was asked

The association between the use of indifference options and the propensity to agree with debrief statement 2 is statistically significant (chi-squared test; forced choice arm vs. indifference arm; p<0.01). Conversely, the association between the wording of the indifference option used and the propensity to agree with debrief statement 2 is not statistically significant (chi-squared test; indifference option 1 arm vs. indifference option 2 arm; p=0.39).

3.7. SENSITIVITY ANALYSIS: EXCLUSIONS LINKED TO DATA QUALITY

Two potential flags of poor data quality or lack of respondent engagement were identified:

1. Choosing the ‘dominated option’ in scenario S7 – i.e. choosing to treat the patient who would gain a life extension of six months from treatment rather than the (otherwise identical) patient who would gain a life extension of 12 months from treatment – even after being given the opportunity to expression indifference

2. Completing the survey in less than 423 seconds – i.e. less than half of the median time taken amongst respondents who had not already been excluded for speeding

One-hundred and ninety respondents (7.9%) chose the dominated option in S7 and therefore hit flag 1. Two-hundred and twenty-one respondents (9.2%) completed the survey in less than 423 seconds and therefore hit flag 2. Twenty-two respondents (0.9%) hit both flags.

Respondents hitting flags 1 or 2 chose to treat the end of life patient (A) more often than respondents who did not (Table 23). Excluding these respondents would therefore *strengthen* the finding that giving priority to the treatment of end of life patients is not supported (Table 24).

Table 23. S1 choices made by all respondents and those who hit data quality flags

	All respondents	Respondents hitting flag 1	Respondents hitting flag 2
A	31.9%	54.7%	36.7%
I	34.7%	20.5%	36.2%
B	33.4%	24.7%	27.1%

Table 24. S1 choices after exclusions based on flags 1 and 2

	No exclusions	Exclude respondents hitting flag 1	Exclude respondents hitting flag 2
A	31.9%	29.9%	31.4%
I	34.7%	35.9%	34.5%
B	33.4%	34.2%	34.1%

3.8. ATTITUDINAL QUESTIONS

Responses to the six attitudinal questions are shown in Table 25. Overall, respondents showed a tendency to agree with the statements presented, with the ‘strongly disagree’ or ‘moderately disagree’ responses accounting for only 13.7% of all responses made across the six statements. Sizeable minorities of respondents agreed with multiple statements that appear, *prima facie*, to describe competing and non-concordant priority-setting objectives: 587 respondents (24.4%) agreed – either strongly or moderately – with attitudinal statements 1, 2 *and* 3; while 866 respondents (36.1%) agreed – either strongly or moderately – with attitudinal statements 4, 5 *and* 6.

Table 25. Responses to attitudinal questions

The health service should:	Agree strongly	Agree moderately	Neither agree not disagree	Disagree moderately	Disagree strongly	Total
1. give priority to extending the life of patients who are expected to die soon as a result of a medical condition	308 (12.8%)	752 (31.3%)	801 (33.4%)	434 (18.1%)	106 (4.4%)	2,401 (100.0%)
2. give priority to treating patients who will get the largest amount of benefit from treatment	757 (31.5%)	982 (40.9%)	484 (20.2%)	131 (5.5%)	47 (2.0%)	2,401 (100.0%)
3. give the same priority to treating all patients, regardless of how ill they are or when they will die	792 (33.0%)	582 (24.2%)	622 (25.9%)	346 (14.4%)	59 (2.5%)	2,401 (100.0%)
4. give priority to improving the quality of life of patients who are expected to die soon as a result of a medical condition	551 (22.9%)	903 (37.6%)	685 (28.5%)	216 (9.0%)	46 (1.9%)	2,401 (100.0%)
5. give priority to extending the life of patients who are expected to die soon as a result of a medical condition	405 (16.9%)	824 (34.3%)	771 (32.1%)	332 (13.8%)	69 (2.9%)	2,401 (100.0%)
6. give equal priority to improving the quality of life and extending the life of patients who are expected to die soon as a result of a medical condition	839 (34.9%)	789 (32.9%)	591 (24.6%)	161 (6.7%)	21 (0.9%)	2,401 (100.0%)

Statements 1 and 5 are identical: the former was presented alongside statements 2 and 3, whereas the latter was presented alongside statements 4 and 6. Just over half of the respondents (55.4%) indicated exactly the same level of agreement with both statements. Conversely, 236 respondents (9.8%) strongly or moderately *agreed* with statement 1 whilst strongly or moderately *disagreeing* with statement 5, or *vice versa*.

Choosing to treat patient A (the end of life patient) in S1 and agreeing with statement 1 (‘The health service should give priority to extending the life of patients who are expected to die soon as a result of a medical condition’) may both be interpreted as indicators of support for an end of life premium.

Table 26 shows that a slight majority of respondents (50.5%) who chose to treat patient A in S1 did indeed express agreement with statement 1, though a sizeable minority (22.6%) disagreed. Amongst the respondents who chose to treat the non-end of life patient (despite being given an opportunity to express indifference), the most common response to attitudinal statement 1 was to neither agree nor disagree. Indeed, these respondents were more likely to agree than to disagree with statement 1.

Table 26. Responses to attitudinal questions

	Statement 1 ("priority to extending the life of patients who are expected to die soon …") response					
S1 response	Agree strongly	Agree moderately	Neither agree not disagree	Disagree moderately	Disagree strongly	Total
A (end of life)	104 (13.6%)	290 (37.9%)	198 (25.9%)	137 (17.9%)	36 (4.7%)	765 (100.0%)
I	107 (12.8%)	229 (27.5%)	356 (42.7%)	111 (13.3%)	30 (3.6%)	833 (100.0%)
B (non-end of life)	97 (12.1%)	233 (29.0%)	247 (30.8%)	186 (23.2%)	40 (5.0%)	803 (100.0%)

In principle, choosing A (quality of life-improving treatment), B (life-extending treatment) or I (indifference option) in S4 would be consistent with agreeing with statements 4, 5 or 6, respectively. In fact, Table 27, Table 28 and Table 29 show that many respondents did not agree with the statement aligned to their choice in S4, in spite of the overall tendency to express agreement with all attitudinal statements.

Table 27. Cross-tabulation: S4 vs. attitudinal statement 4

	Statement 4 ("priority to improving the quality of life …") response					
S4 response	Agree strongly	Agree moderately	Neither agree not disagree	Disagree moderately	Disagree strongly	Total
A (quality of life gain)	248 (25.6%)	391 (40.4%)	220 (22.7%)	91 (9.4%)	19 (2.0%)	969 (100.0%)
I	177 (21.7%)	256 (31.3%)	303 (37.1%)	67 (8.2%)	14 (1.7%)	817 (100.0%)
B (life extension)	126 (20.5%)	256 (41.6%)	162 (26.3%)	58 (9.4%)	13 (2.1%)	615 (100.0%)

Table 28. Cross-tabulation: S4 vs. attitudinal statement 5

	Statement 5 ("priority to extending the life …") response					
S4 response	Agree strongly	Agree moderately	Neither agree not disagree	Disagree moderately	Disagree strongly	Total
A (quality of life gain)	138 (14.2%)	337 (34.8%)	283 (29.2%)	179 (18.5%)	32 (3.3%)	969 (100.0%)
I	405 (17.1%)	251 (30.7%)	318 (38.9%)	84 (10.3%)	24 (2.9%)	817 (100.0%)
B (life extension)	127 (20.7%)	236 (38.4%)	170 (27.6%)	69 (11.2%)	13 (2.1%)	615 (100.0%)

Table 29. Cross-tabulation: S4 vs. attitudinal statement 6

	Statement 6 ("equal priority to improving the quality of life and extending the life …") response					
S4 response	Agree strongly	Agree moderately	Neither agree not disagree	Disagree moderately	Disagree strongly	Total
A (quality of life gain)	329 (34.0%)	338 (34.9%)	213 (22.0%)	77 (7.9%)	12 (1.2%)	969 (100.0%)

	Statement 6 (“equal priority to improving the quality of life and extending the life …”) response					
S4 response	Agree strongly	Agree moderately	Neither agree not disagree	Disagree moderately	Disagree strongly	Total
I	272 (33.3%)	260 (31.8%)	234 (28.6%)	47 (5.8%)	4 (0.5%)	817 (100.0%)
B (life extension)	238 (38.7%)	191 (31.1%)	144 (23.4%)	37 (6.0%)	5 (0.8%)	615 (100.0%)

Levels of internal incoherence – that is, providing responses to attitudinal questions that appear at odds with one’s earlier responses to the choice tasks – did not vary much between study arms. For example, the proportion of respondents who chose to treat the end of life patient in S1 whilst agreeing with attitudinal statement 1 ranged from 50.3% in the forced choice arm to 52.3% in the indifference arm; and from 50.0% in the no visual aid arm to 53.0% in the visual aid arm.

3.9. IMPACT OF RESPONDENT BACKGROUND CHARACTERISTICS IN S1

The results of the multiple linear regression are shown in Table 30. Three background characteristics were found to have coefficients that were statistically significant at the 5% level: age, children and experience of terminal illness. Respondents who are younger, have responsibility for children and have experience of terminal illness were more likely than average to choose to treat the end of life patient. However, when considering the subgroup of respondents meeting all three criteria (i.e. respondents who are younger than the median age of 47 years *and* have responsibility for children *and* have experience of terminal illness; n=326), the majority (60.1%) did *not* choose to treat the end of life patient.

Table 30. Impact of background characteristics – results of regression

Variable	Coefficient	Standard error	p-value
Age	-0.0076	0.0033	0.020
Gender	0.0578	0.0955	0.545
Social grade	0.0813	0.0958	0.396
Children	0.2859	0.1025	0.005
Degree	-0.0230	0.0950	0.809
Health limitations	0.1172	0.0711	0.099
Experience of terminal illness	0.2777	0.0989	0.005
Forced choice arm	0.5060	0.0935	0.000
Visual aid arm	0.0135	0.0903	0.882
Constant	-1.0391	0.2607	0.000

Observations (n): 2,316
Log-likelihood: -1421.2 (LR test: chi-squared = 58.1; degrees of freedom = 9; p<0.01)
Pseudo-R^2: 0.020

3.10. ORDERING EFFECTS

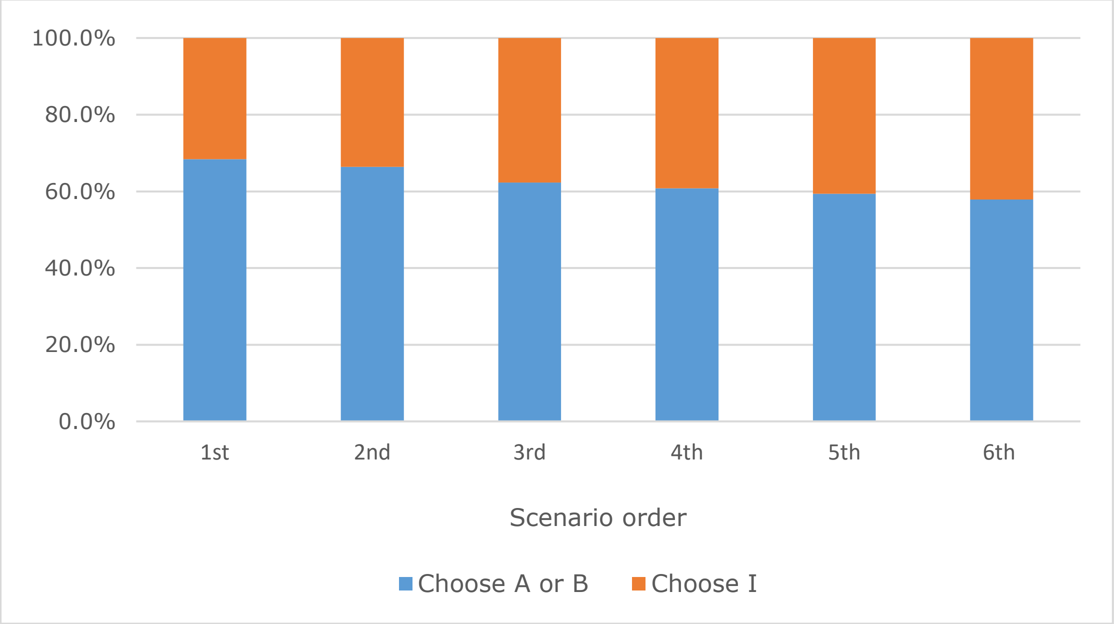
The order in which scenarios S1 to S6 were presented was randomised for each respondent. Table 31 shows how the responses made differed depending on whether or not the scenario in question was the first to be presented. In each column, the modal choice is emboldened. In S1 and S3, the modal choice when those scenarios were presented first differs from the modal choice when they were presented later. In all six scenarios, indifference was expressed less often when the scenario was presented first. Pooling responses from the six scenarios, the association between scenario ordering (first or not first) and the propensity to express indifference is statistically significant (chi-squared test; $p<0.01$).

Table 31. Aggregate S1 to S6 responses, split by whether scenario appeared first or not

Scenario	S1		S2		S3		S4		S5		S6	
Ordering	First	Not first	First	Not first	First	Not first	First	Not first	First	Not first	First	Not first
A	**141 (37.0%)**	624 (30.9%)	50 (12.5%)	290 (14.5%)	97 (25.7%)	482 (23.8%)	**164 (41.0%)**	**805 (40.2%)**	**168 (38.5%)**	**756 (38.5%)**	**181 (44.5%)**	**843 (42.3%)**
I	125 (32.8%)	**708 (35.1%)**	100 (25.1%)	523 (26.1%)	130 (34.4%)	**784 (38.8%)**	107 (26.8%)	710 (35.5%)	113 (25.9%)	594 (30.2%)	128 (31.5%)	763 (38.3%)
B	115 (30.2%)	688 (34.1%)	**249 (62.4%)**	**1,189 (59.4%)**	**151 (40.0%)**	757 (37.4%)	129 (32.3%)	486 (24.3%)	155 (35.6%)	615 (31.3%)	98 (24.1%)	388 (19.5%)
Total	381 (100.0%)	2,020 (100.0%)	399 (100.0%)	2,002 (100.0%)	378 (100.0%)	2,023 (100.0%)	400 (100.0%)	2,001 (100.0%)	436 (100.0%)	1,965 (100.0%)	407 (100.0%)	1,994 (100.0%)

Respondents in the indifference arm were increasingly likely to express indifference in the initial question of each scenario (thereby avoiding the follow-up question) as they proceeded through the survey (Figure 8).

Figure 8. Proportion choosing A or B versus proportion choosing I, by scenario order



4. DISCUSSION

This study used an internet survey to elicit the preferences of a large sample of the general public, representative in terms of age, gender and social grade, regarding the prioritisation of treatments for patients with short life expectancy. Nine hypotheses were tested. Some of these hypotheses relate closely to NICE's supplementary end of life policy, which in effect involves placing greater weight on a unit of health gain generated by life-extending end of life treatments than on that generated by other types of treatments (regardless of how old the patients in question are or for how long they have known about their prognosis). Other hypotheses were tested in order to examine methodological issues, such as whether people's preferences regarding an end of life premium are affected by the inclusion of visual aids or an indifference option in the survey used to elicit those preferences. The results relating to each hypothesis are discussed below.

The most straightforward test of public support for an end of life premium (hypothesis 1) was in scenario S1, in which respondents were asked to choose between giving a life extension to a patient with one year of life expectancy and an equal-sized life extension to another patient with five years of life expectancy. Responses were very evenly split across the three available options – the most common accounted for less than 35% of all responses. Overall, choosing to treat the end of life patient was the *least* popular choice, though considerable variation in response patterns across arms can be observed – for respondents in the forced choice arm, for example, choosing to treat the end of life patient was the *most* popular choice. Nevertheless, in none of the study arms did a majority of respondents choose to treat the end of life patient. Hence, it can be concluded that the results observed are not consistent with an end of life premium.

The results for scenario S2 were clearer and more robust than those for the other scenarios. The majority of respondents chose to treat a younger non-end of life patient rather than an older end of life patient. This result was observed in all study arms, and is consistent with findings elsewhere that people become less concerned about patients' remaining life years when those patients are relatively old[23-25]. NICE's general principle is that health care resources cannot be allocated or restricted on the basis of age[26-28]. Nevertheless, it is still informative that people's preferences regarding end of life appear to be context-specific, and specifically that preferences regarding an end of life premium seem to be affected by the ages of the patients in question (hypothesis 2). The finding is particularly pertinent if the treatments meeting NICE's end of life criteria tend to target older patients.

Although the shifts in response patterns between scenarios S1 and S3 were modest in comparison to other pairs of scenarios, the results indicate that people's preferences regarding an end of life premium appear to be affected by how long the patients in question have known about their prognosis (hypothesis 3). Respondents were more likely to choose to treat the patient with shorter life expectancy when advised that the patient had just been diagnosed, as opposed to having been diagnosed five years ago. A limitation of the design of S3 is that by the patients' expected times of death without treatment, the difference in how long they would have known about their prognosis is small – patient A would have known about their prognosis for six years; patient B for five years. The effect of time with knowledge might have been stronger had there been a greater discrepancy between the situations facing the two patients, though there is a risk that the resulting scenario would have been considered implausible by respondents.

Scenario S4 is of interest because NICE's policy accommodates life-extending but not quality of life-improving end of life treatments. By contrast, the majority of respondents in this survey did *not* express preference for providing the life-extending end of life treatment (hypothesis 4). Indeed, the proportion of respondents choosing the quality of life-improving treatment exceeded the proportion choosing the life-extending treatment in all study arms. Hence, the responses to S1 and S4 suggest that public support for NICE's end of life policy is limited. The preference for quality of life improvements is further demonstrated by the responses to scenario S5. In that scenario, where the end of life treatment was quality of life-improving, respondents were more likely to choose to treat the end of life patient than in S1, where the end of life treatment was life-extending (hypothesis 5).

However, the responses to S4 should be considered alongside those of S6, which suggest that the preference of quality of life improvements over life extensions is not specific to the end of life context (hypothesis 6). As noted above, respondents were advised that patients consider living in 50% quality of life for two years to be equally desirable as living in 100% quality of life for one year. An implication of this assumption (albeit not explained explicitly to respondents in this way) is that a 50% quality of life improvement lasting 12 months is equally desirable to patients as a 12-month life extension at 50% quality of life. This means that the (undiscounted) gains generated by treating patient A are equivalent to those generated by treating patient B in S4, S5 and S6. Provided that the respondents understood and accepted the information given, the fact that they were more likely to choose the quality of life-improving option in these scenarios appears to indicate that they consider a quality of life improvement worth half a QALY to be more socially valuable than a life extension worth half a QALY. Another possibility is that respondents ignored or rejected the information provided about the patients' preferences, assuming instead that the patients would prefer the quality of life improvement for themselves.

One way of disentangling individual and social values would be to design a study which asks respondents to provide their values for defined states of quality of life from an individual perspective (for example, using time trade-off) and then to evaluate those same states from a social decision-maker perspective, using those values. See Dolan and Green[29] for an example of how a study comparing differences between individual and social values might be designed, though it should be noted that the methods used in that study would be challenging to apply in a self-completion internet survey.

Instead, this study attempted to explore the effect of perspective on preferences for an end of life premium (hypothesis 7) by including scenario S8, an explicitly individual perspective operationalisation of S1. The results show that respondents were considerably less likely to prioritise the provision of the end of life treatment when answering from an individual perspective. This finding can be contrasted to those of the individual perspective studies included in the review by Shah *et al*.[3]. Those studies, all of which used the willingness-to-pay method, reported relatively high values for health gains in end of life scenarios (with the exception of Shiroiwa *et al.*[30]). It may be that by presenting the end of life and non-end of life scenarios as two possible states of the world each with a 50% chance of occurring, and by removing explicit consideration of money from the tasks, this study managed to overcome some of the features of willingness-to-pay studies that render them potentially unsuitable for informing society-level decision-making. It is acknowledged that S8 may be considered the most abstract and contrived of the scenarios presented in the survey, though all of the pilot respondents claimed that it was easy to understand.

The observation in the literature review that studies that include visual aids appear more likely than average to report evidence consistent with an end of life premium[3] was not repeated in this study. In four of the scenarios, including S1, the effect of the visual aid was modest. In the three scenarios in which the choice was between a quality of life improvement and a life extension (S4, S5, S6), the quality of life improvement was chosen less frequently when a visual aid was used. It seems therefore that visual presentation of information can have an impact on people's choices about priority-setting, particularly when presenting information about quality of life – a concept that can be difficult for some people to comprehend.

In all scenarios, respondents in the no visual aid arm expressed indifference more often than those in the visual aid arm, potentially implying that the visual aids helped respondents to distinguish between the alternatives and to be more decisive (though this conjecture is not supported by the responses to the debrief questions, which did not differ greatly between the visual aid and no visual aid arms). Respondents in the visual aid arm spent longer than average completing the survey, presumably because there was more information on the screen to make sense of.

It should be noted that the visual aids used in this study were similar to those used in other studies of end of life-related preferences (for example, [6, 14, 28, 31]), relying on conceptual diagrams with quality of life depicted on the vertical axis and length of life on the horizontal axis. This is not the only form that visual aids to support priority-setting scenarios can take. For example, in a group discussion study investigating public support for various ethical principles of health care rationing, Cookson and Dolan[32] used photographs of actors to represent hypothetical patients. The observed effect of conceptual diagrams in this study cannot be generalised to other, substantially different forms of visual aids.

Compared to visual aids, the effect of including an explicit indifference option was less ambiguous. In S1, the modal choice amongst respondents in the forced choice arm – choosing to treat the end of life patient – was the *least* common choice for respondents in the indifference arm. Although this option was never chosen by a majority of respondents in any of the study arms, it is clear that the balance of responses to the choice tasks was influenced by what response options were available. In most of the scenarios, respondents were more likely to express indifference when an indifference option was offered in the initial question than when indifference could only be expressed indirectly via the follow-up question.

The results provide evidence that it is not only *whether* an indifference option is available that matters, but also *how* exactly that indifference option is framed and mechanised. In this study, respondents were on the whole more likely to choose the option worded 'Both patients should have an equal chance of being treated (tossing a coin would be a fair way to make the choice)' than the option worded 'I have no preference (I do not mind which patient is treated)'. This is in spite of the fact that both statements have identical implications for the allocation of health care resources. Alternative indifference options, such as 'I am not able to make a decision and would prefer that the choice be made by others'[33], were considered but not included in the design in order to reduce complexity, and may well have generated different results.

It is acknowledged that the follow-up questions used in this study were complicated, even after efforts to simplify the wording of the instructions following the pilot. Respondents in the forced choice arm were always made to answer the follow-up question, whereas respondents in the indifference arm only answered the follow-up question when they did not choose the indifference option in the initial question. This may be reflected in respondents' responses to debrief statement 2 ('It was difficult to understand the questions I was asked'). Respondents in the indifference arm were more likely than those in the forced choice arm to disagree with this statement. This was particularly true of the 130 respondents who *always* expressed indifference in the initial question (and therefore never proceeded to the follow-up question) – disagreeing strongly with debrief statement 2 was the modal response (35%) amongst this group.

Further, a plausible explanation of the finding that respondents in the indifference arm were increasingly likely to choose the indifference option as they proceeded through the survey is that they learned that this was how to avoid the follow-up question, and therefore to reduce the time and effort needed to complete the survey. For surveys including multiple questions that are due to be compared with each other, randomisation of question order across respondents can help to minimise the impact of order bias, though the most appropriate specific randomisation mechanism is likely to vary from study to study. At the very least, studies should report whether or not the question order was randomised.

The follow-up question in scenarios S1 to S7 involved asking respondents to specify a size of gain that would make them indifferent between treating patient A and patient B. This mechanism was similar to that used by Abel Olsen[15], and chosen based on a judgement that it would generate more information than alternative approaches. The results indicate that when faced with a large number of response options, most respondents tend to be drawn to a small subset of those options (such as the mid-point value), implying the use of simplifying heuristics. An alternative approach, as used by Linley and Hughes[34], would be to repeat the initial question but with a reduced size of gain for the patient that was preferred initially. This would generate information about the strength of respondents' preferences, but would not in itself identify their points of indifference. An extension to this approach would be to apply a specified procedure of choice iterations to guide the respondent towards their point of indifference (i.e. by repeatedly increasing or reducing the size of gain for a given patient depending on the respondent's previous choice). Such iterative procedures are commonly used in the application of the standard gamble, person trade-off, time trade-off and willingness-to-pay techniques, though they are themselves also a potential source of biases and heuristics[35, 36].

The attitudinal questions were included as an alternative means of capturing respondents' views on priority-setting. Fewer than half (44.1%) of the respondents agreed with the statement that 'The health service should give priority to extending the life of patients who are expected to die soon as a result of a medical condition'. This is somewhat greater than the 31.9% of respondents who chose to treat the end of life patient in S1, but still represents the lowest level of support observed across all six statements. The discrepancy may reflect respondents' interpretation of the statement – they may have assumed a life extension greater than and/or a life expectancy without treatment shorter than the ones presented in S1. An overall tendency to express agreement with the statements can be observed, with many respondents agreeing with statements that appear, *prima facie*, to be inconsistent with their responses to the choice tasks and to other statements describing competing priority-setting objectives – for example, agreeing with statements 4, 5 *and* 6. In contrast to the results of this study, Rowen *et al.*[18] reported that a lower level of support for an end of life premium could be discerned from respondents' responses to attitudinal questions than from their responses to the preceding choice (DCE) tasks.

It should be noted that although the attitudinal questions were designed so as to align with certain responses to the scenario questions, the type types of questions are not perfectly consistent with each other. For example, a respondent could have chosen to treat the end of life patient in scenario S1 whilst disagreeing with attitudinal statement 1 because they saw attitudinal statement 1 being presented alongside attitudinal statement 2 ('The health service should give priority to treating patients who will get the largest amount of benefit from treatment') and agreed more with attitudinal statement 2 than with attitudinal statement 1. In S1, both patients would get the same amount of benefit from treatment so respondents could not make a choice that involved one patient getting a larger benefit. However, it is trickier to think of a coherent reason why a respondent would choose to treat the *non*-end of life patient in S1 whilst *agreeing* with attitudinal statement 1, which 41.1% of respondents did. Ultimately, the only way to have perfectly matched the attitudinal statements with the scenario questions would have been to add caveats and nuances to the statements. This may defeat the purpose of the exercise given that the intention of the attitudinal questions was to offer a more general and less convoluted alternative to the scenario-based choice tasks.

The fact that 36.1% of respondents agreed with three statements that were intended to be mutually exclusive from each other (4, 5 and 6) suggests that the responses may have been distorted by acquiescence bias[37] – that is, the tendency to agree when in doubt – and casts doubt on the usefulness of this type of exercise. This issue is avoided in choice tasks such as the initial questions in the scenarios in this study, which specified that only one of the two patients could be treated and therefore required sacrifices to be made. A potential solution would be to ask respondents to indicate which of multiple competing attitudinal statements they agreed with most, thereby forcing them to prioritise amongst several policy statements that they are inclined to agree with.

Some limitations of the study should be mentioned. Whilst the study design and analyses undertaken are deemed to be suitable for testing the hypotheses set out in 6.1**Error! Reference source not found.**, it is acknowledged that in many cases alternative approaches could have been followed. For example, hypothesis 5 was examined by testing for an association between choosing a life-extending end of life treatment over a life-extending non-end of life treatment (in S1) and choosing a *quality of life-improving* end of life treatment over a life-extending non-end of life treatment (in S5). It is unclear whether the same result would have been achieved if the treatment for the non-end of life patient in both scenarios had been quality of life-improving rather than life-extending. This would have been an alternative, and legitimate, means of testing hypothesis 5.

Most of the analyses undertaken focused on the propensity to choose to treat patient A in each of the scenarios (in S1, S2 and S3, patient A was the end of life patient; in S4, S5 and S6, patient A stood to receive a quality of life-improving treatment). This meant that the analyses were largely binomial (i.e. A versus I/B) in nature. Given that the study was to a large extent motivated by questions about public support for NICE's end of life policy, this focus seems reasonable. However, the conclusions made about the impact of scenario information, experimental modifications and other explanatory factors might have been different if the analyses had instead been multinomial (i.e. A versus I versus B). For the purpose of informing the design of stated preference studies, it may be just as useful to understand what drives people to express indifference in favour of choosing to treat patient B, and *vice versa*. However, this level of analysis was considered to be beyond the scope of the study.

The study design involved adjusting a single factor or attribute (such as time with knowledge) from one scenario or arm to another, and using cross-tabulations to analyse the impact of that attribute on choices. This allowed the impact of a large number of attributes to be isolated to a greater degree than might have been possible using other methods such as DCE. However, only a small number (usually two) of levels for any given attribute were tested – for example, time with knowledge was set to either zero or five years, and the data are insufficient to make claims about the effect that other possible levels might have had. Further, the isolation of changes between scenarios may have resulted in a focusing effect whereby the importance of the varying attribute was exaggerated, though the randomisation of scenario ordering should have mitigated this effect to an extent. It has been suggested that seeking choices between packages of attributes that vary in multiple ways lessens such focusing effects and makes it more difficult for respondents to answer strategically[38].

The level of drop-out from the survey (32.6%; comprising individuals who were screened in but either did not consent to take part or did not complete the survey) was higher than expected. It is unclear whether the high drop-out rate was due to respondent fatigue, technical problems, or some other cause. A drawback associated with internet surveys is that they offer limited opportunities to investigate reasons why respondents fail to complete the survey or give responses that seem internally incoherent or contrary to researchers' expectations (though no major issues with the present survey were identified in the face-to-face interviews conducted as part of the pilot). Further limitations of internet surveys are discussed elsewhere[6].

Notwithstanding the caveats and limitations described above, this study has addressed some of the gaps in the empirical literature on public preferences regarding the social value of end of life treatments[3]. Little support for NICE's end of life policy is observed, with the majority of respondents rejecting the opportunity to prioritise the treatment of end of life patients over non-end of life patients, or to provide life-extending end of life treatments over quality of life-improving treatments. Specifying that the end of life patient is relatively old or has known about their prognosis for some time, or asking the questions from an individual rather than a social decision-maker perspective, weakens further the evidence of support for an end of life premium. Study design considerations – specifically, the use of visual aids and the availability of explicit indifference options – were found to affect respondents' choices, though in no version of the survey was a majority preference for treating the end of life patient observed.

A finding of potential interest is that the responses to the choice tasks indicate that support for NICE’s end of life policy (or some variant of it) is stronger than support for a pure QALY-maximisation approach to health care priority-setting. But a more noteworthy finding is that very few respondents (less than 4%) made choices that imply unambiguous support for either QALY-maximisation or for (even a less prescriptive version of) NICE’s end of life policy.

5. CONCLUSIONS

This paper has described a large-scale study that examined the extent of public support for an end of life premium, and the impact of study design considerations and framing effects on end of life-related preferences. The results are not consistent with an end of life premium – *ceteris paribus*, only a minority of respondents chose to give priority to the end of life patient over the non-end of life patient. This minority was reduced further when the end of life patient was described as older than and/or as having known about their prognosis for longer than the non-end of life patient. The use of an individual (rather than a social decision-maker) perspective also further weakened the case for an end of life premium. A preference for quality of life improvements over life extensions (holding the size of QALY gain constant) was observed, though this preference does not appear to be specific to the end of life context and was elicited under a social decision-maker perspective only. A caveat to these results is that a number of discrepancies were found between respondents' responses to the choice tasks and their subsequent responses to the attitudinal questions. The study adds not only to the evidence on end of life-related preferences but also to the evidence on framing effects in stated preference research. Respondents' choices were found to be sensitive to the inclusion of indifference options and (to a lesser extent) visual aids.

6. REFERENCES

1. Rawlins, M., Barnett, D. and Stevens, A., *Pharmacoeconomics: NICE's approach to decision-making.* British Journal of Clinical Pharmacology, 2010. **70(3)**: p. 346-349.
2. National Institute for Health and Care Excellence, *Appraising life-extending, end of life treatments.* NICE London: National Institute for Health and Care Excellence., 2009.
3. Shah, K.K., Tsuchiya, A. and Wailoo, A.J., *Valuing health at the end of life: a review of stated preference studies in the social sciences literature. .* Social Science & Medicine, 2018. **204**: p. 39-50.
4. Shah, K., Wailoo, A. and Tsuchiya, A., *Valuing health at the end of life: an exploratory preference elicitation study. .* 2011.
5. Shah, K.K., Tsuchiya, A. and Wailoo, A.J., *Valuing health at the end of life: an empirical study of public preferences.* European Journal of Health Economics, 2014. **15(4)**: p. 389-399.
6. Shah, K.K., Tsuchiya, A. and Wailoo, A.J., *Valuing health at the end of life: a stated preference discrete choice experiment.* Social Science and Medicine, 2015a. **124**: p. 48-56.
7. Cookson, R., *Can the NICE “end-of-life premium” be given a coherent ethical justification? .* Journal of Health Politics, Policy and Law, 2013. **38(6)**: p. 1129-1148.
8. McHugh, N., Baker, R.M., Mason, H., Williamson, L., van Exel, J., Deogaonkar, R., Collins, M. and Donaldson, C., *Extending life for people with a terminal illness: a moral right and an expensive death? Exploring societal perspectives.* BMC Medical Ethics, 2015. **16(14)**.
9. Dolan, P., Olsen, J.A., Menzel, P. and Richardson, J. , *An inquiry into the different perspectives that can be used when eliciting preferences in health.* Health Economics, 2003. **12(7)**: p. 545-551.
10. Tsuchiya, A., and Watson, V., *Re-thinking ‘The different perspectives that can be used when eliciting preferences in health’.* Health Economics, 2017. **26(12)**: p. e103-e107.
11. Plous, S., *The psychology of judgment and decision making.* New York: McGraw-Hill, 1993.
12. Luchini, S., and Watson, V., *Uncertainty and framing in a valuation task.* Journal of Economic Psychology, 2013. **39**: p. 204-214.
13. Mason, H., Baker, R. and Donaldson, C., *Understanding public preferences for prioritizing health care interventions in England: does the type of health gain matter?* Journal of Health Services Research and Policy, 2011. **16(2)**: p. 81-89.
14. Rowen, D., Brazier, J., Mukuria, C., Keetharuth, A, Risa Hole, A., Tsuchiya, A., Whyte, S. and Shackley, P., *Eliciting societal preferences for weighting QALYs for burden of illness and end of life.* Medical Decision Making, 2016a. **36(2)**: p. 210-222.
15. Abel Olsen, J., *Priority preferences: “end of life” does not matter, but total life does.* Value in Health, 2013. **16(6)**: p. 1063-1066.
16. Cummings, R.G., and Taylor, L.O., *Unbiased value estimates for environmental goods: a cheap talk design for the contingent valuation method.* The American Economic Review, 1999. **89(3)**: p. 649-665.

17. Özdemir, S., Johnson, F.R. and Hauber, A.B., *Hypothetical bias, cheap talk, and stated willingness to pay for health care.* Journal of Health Economics, 2009. **28(4)**: p. 894-901.
18. Rowen, D., Brazier, J., Mukuria, C., Keetharuth, A., Hole, A.R., Tsuchiya, A., Whyte, S. and Shackley, P., *Eliciting societal preferences for weighting QALYs for burden of illness, size of gain and end of life.* 2014.
19. Shah, K., Chapman, A., Devlin, N. and Barnsley, P., *Do respondents completing abstract, hypothetical priority-setting exercises agree with the policy implications of their choices?* 2015b.
20. StataCorp, 2013.
21. Office for National Statistics, *2011 Census, Population Estimates by single year of age and sex for Local Authorities in the United Kingdom.* 2011(Available from: http://www.ons.gov.uk/ [accessed 23 Oct 2014]).
22. National Readership Survey, *Social Grade. Available at: http://www.nrs.co.uk/nrs-print/lifestyle-and-classification-data/social-grade/.* 2015.
23. Dolan, P., and Shaw, R., *A note on a discussion group study of public preferences regarding priorities in the allocation of donor kidneys.* Health Policy, 2004. **68(1)**: p. 31-36.
24. Stahl, J.E., Tramontano, A.C., Swan, J.S. and Cohen, B.J., *Balancing urgency, age and quality of life in organ allocation decisions—what would you do?: a survey.* Journal of Medical Ethics, 2008. **34(2)**: p. 109-115.
25. Richardson, J., Sinha, K., Iezzi, A. and Maxwell, A., *Maximising health versus sharing: Measuring preferences for the allocation of the health budget.* Social Science and Medicine, 2012. **75(8)**: p. 1351-1361.
26. National Institute for Health and Care Excellence, *Social value judgements: principles for the development of NICE guidance, 2nd edition.* NICE London: National Institute for Health and Care Excellence., 2008c.
27. Shah, K.K., Cookson, R., Culyer, A.C. and Littlejohns, P., *NICE's social value judgements about equity in health and health care.* Health Economics, Policy and Law, 2013a. **8(2)**: p. 145-165.
28. Rowen, D., Brazier, J., Keetharuth, A., Tsuchiya, A. and Mukuria, C., *Comparison of modes of administration and alternative formats for eliciting societal preferences for burden of illness Comparison of modes of administration and alternative formats for eliciting societal preferences for burden of illness. .* Applied Health Economics and Health Policy, 2016b. **14(1)**: p. 89-104.
29. Dolan, P., and Green, C., *Using the person trade-off approach to examine differences between individual and social values.* Health Economics, 1998. **7(4)**: p. 307-322.
30. Shiroiwa, T., Igarashi, A., Fukuda, T. and Ikeda, S., *WTP for a QALY and health states: more money for severer health states?* Cost Effectiveness and Resource Allocation, 2013. **11(22)**.
31. Pennington, M., Baker, R., Brouwer, W., Mason, H., Hansen, D. G., Robinson, A. and Donaldson, C., *Comparing WTP values of different types of QALY gain elicited from the general public.* Health Economics, 2015. **24(3)**: p. 280-293.
32. Cookson, R., and Dolan, P., *Public views on health care rationing: a group discussion study.* Health Policy, 1999. **49(1-2)**: p. 63-74.

33. Green, C., *Investigating public preferences on 'severity of health' as a relevant condition for setting healthcare priorities.* Social Science and Medicine,, 2009. **68(12)**: p. 2247-2255.
34. Linley, W.G., and Hughes, D.A., *Societal views on NICE, Cancer drugs fund and value-based pricing criteria for prioritising medicines: a cross-sectional survey of 4118 adults in Great Britain.* Health Economics,, 2013. **22(8)**: p. 948-964.
35. Ternent, L., and Tsuchiya, A., *A note on the expected biases in conventional iterative health state valuation protocols.* Medical Decision Making, 2013. **33(4)**: p. 544-546.
36. Oppe, M., Rand-Hendriksen, K., Shah, K., Ramos-Goni, J.M. and Luo, N., *EuroQol protocols for time trade-off valuations of health outcomes.* Pharmacoeconomics, 2016. **34(10)**: p. 993-1004.
37. Messick, S.J., *The psychology of acquiescence: an interpretation of research evidence.* In: Berg, I.A., ed. Response set in personality assessment. Chicago: Aldine., 1967.
38. Sheldon, R., *Stated preference methods for eliciting values.* 1999.