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Valuing health at the end of life

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Abstract

This thesis examines whether members of the public wish to place greater weight on a unit of health gain for end of life patients (i.e. patients with short life expectancy) than on that for other types of patients. The research question was motivated by a policy introduced in 2009 by the National Institute for Health and Care Excellence (NICE). The policy indicates that under certain circumstances, life-extending end of life treatments may be recommended for use in the National Health Service even if they would not normally be considered a cost-effective use of health care resources. NICE's policy was justified in part by claims that it reflected the preferences of society, but little evidence was available to support the premise that society favours such an 'end of life premium'. This thesis helps to fill the gap in the evidence.

Four empirical studies were undertaken, each using hypothetical choice exercises to elicit the stated preferences of the UK general public regarding the value of health gains for end of life patients (total n=6,441). A variety of preference elicitation techniques, modes of administration and analytical approaches were used. Results varied across studies, but overall the evidence is not consistent with an end of life premium. Whereas NICE's end of life policy applies to life-extending treatments, there is some evidence that quality of life improvements are more highly valued than life extensions for end of life patients. The results of all four studies suggest that where a preference for prioritising the treatment of end of life patients does exist, this preference may be driven by concerns about how long the patients have known about their prognosis rather than how long they have left to live *per se*. End of life-related preferences also appear to be sensitive to framing effects and study design choices.

Author's declaration

I declare that this thesis is my original work. No part of this work has previously been submitted for a degree at another institution. All substantial sources of assistance have been acknowledged. All external sources of information are cited as appropriate.

My supervisors, Professor Aki Tsuchiya and Professor Allan Wailoo, provided guidance and critical review throughout my doctoral studies. They contributed to the design and interpretation of the studies reported in Chapter 2, 3, 4, 5 and 6, and should be considered secondary co-authors of those chapters. I held primary responsibility for all aspects of the research in this thesis, and should be considered the lead author of Chapters 2, 3, 4, 5 and 6, and the sole author of Chapters 1 and 7. Dr Arne Risa Hole provided substantial input for part of the study reported in Chapter 5. He used the Stata software package to construct an experimental design according to the requirements I gave to him. He also provided advice on data analysis and interpretation. Once I had received this advice I conducted the various analyses unassisted. Dr Hole provided comments on an earlier draft version of the study write-up and at all times confirmed that he was happy with my understanding of the methods being used and with my interpretation of the results.

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Prior publications and presentations

Publications

Selected material in Chapter 1 has been published in *Health Economics, Policy and Law* (Shah *et al.*, 2013a).

A previous version of Chapter 2 was presented and discussed at the January 2016 Health Economists' Study Group meeting (discussant: Jytte Seested Nielsen). Selected findings from Chapter 2 have also been published as a chapter in the book *Care at the end of life: an economic perspective* (Shah, 2016a).

A previous version of Chapter 3 has been published as an Office of Health Economics Research Paper (Shah *et al.*, 2011) following peer review by Tony Culyer (a member of the Office of Health Economics Editorial Committee).

A previous version of Chapter 4 has been published in *European Journal of Health Economics* (Shah *et al.*, 2014).

A previous version of Chapter 5 has been published in *Social Science and Medicine* (Shah *et al.*, 2015a).

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Presentations

Shah, K., 2016. Discussion of presentation by Baker, R.: "Perspectives, priorities and plurality: Eliciting societal values and the relative value of life extension at the end of life." Oral presentation at the London Health Economics Group meeting. London. 17 November.

Shah, K., 2016. Does society wish to prioritise end-of-life treatments over other types of treatment? Oral presentation at Priorities 2016, 11th International Conference of the International Society on Priorities in Health. Birmingham. 7-9 September.

Shah, K., Tsuchiya, A. and Wailoo, A., 2016. Does society wish to place greater weight on a unit of health gain for end-of-life patients than on that for other types of patients? Poster presentation at the ISPOR Congress. Washington, DC. 21-25 May.

Shah, K., Tsuchiya, A. and Wailoo, A., 2016. Do members of the general 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? Paper presented at the Health Economists' Study Group meeting. University of Manchester. 6-8 January.

Shah, K., 2015. Valuing health at the end of life. Oral presentation at the iBMG Health Economics seminar series. Institute of Health Policy & Management, Erasmus University Rotterdam. 22 September.

Shah, K.K., 2014. Valuing health at the end of life: A stated preference discrete choice experiment. Oral presentation at the HERC seminar series. Health Economics Research Centre, University of Oxford. 21 January.

Shah, K.K., Tsuchiya, A. and Wailoo, A., 2013. Valuing health at the end of life: A stated preference discrete choice experiment. Paper presented at the Nordic Health Economists' Study Group meeting. Oslo. 14–16 August.

Shah, K.K., 2013. Valuing health at the end of life: A stated preference discrete choice experiment. Oral presentation at the CHE seminar series. Centre for Health Economics, University of York. 11 July.

Shah, K.K., Tsuchiya, A. and Wailoo, A., 2013. Valuing health at the end of life: A stated preference discrete choice experiment. Paper presented at the Health Economists' Study Group meeting. University of Warwick. 26–28 June.

Shah, K.K., 2013. Discussion of paper by Kinghorn, P. and Coast, J.: "The value of time at end of life: Anchoring in economic evaluation". Oral presentation at the Health Economists' Study Group meeting. University of Warwick. 26–28 June.

Shah, K.K., 2013. Valuing health at the end of life. Oral presentation at the OHE lunchtime seminar series. Office of Health Economics, London. 26 March.

Shah, K.K., 2012. Valuing health at the end of life: An empirical study of public preferences. Oral presentation at the 34th Annual Meeting of the Society for Medical Decision Making. Phoenix. 17–20 October.

Shah, K.K., 2012. Valuing health at the end of life: An empirical study of public preferences. Oral presentation at Priorities 2012, 9th International Conference of the International Society on Priorities in Health Care. Vancouver. 16–19 September.

Shah, K.K., 2012. Valuing health at the end of life: An empirical public preference study. Oral presentation at the Society for Medical Decision Making European Meeting. Oslo. 10-12 June.

Shah, K.K., 2012. Valuing health at the end of life: An empirical public preference study. Oral presentation at the AUHE seminar series. Academic Unit of Health Economics, University of Leeds. 8 May.

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Shah, K.K., 2011. Valuing health at the end of life: An empirical public preference study. Oral presentation at the HEDS seminar series. School of Health and Related Research, University of Sheffield. 24 November.

Shah, K.K., Tsuchiya, A. and Wailoo, A.J., 2011. Valuing health at the end of life: An exploratory preference elicitation study. Paper presented at the Issues on Rationing in Health Care Conference. London School of Economics. 19-20 September.

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1 INTRODUCTION

1.1 Aims and scope of the research in this thesis

The primary aim of this thesis is to answer the following research question:

> *Do members of the public wish to place greater weight on a unit of health gain for end of life patients (i.e. patients with short life expectancy) than on that for other types of patients?*

This research question was motivated by a policy introduced in 2009 by the National Institute for Health and Care Excellence (NICE), an agency that provides guidance and advice on the use of health technologies in the National Health Service (NHS) in England. The policy effectively gives higher priority to life-extending end of life treatments than to other types of treatments. This thesis seeks to examine the extent to which such a policy is consistent with the preferences of the general public. Throughout the thesis, the NICE end of life policy is used as the framework for the design of the research. However, the issues explored have relevance in all jurisdictions seeking to understand the extent of public support for giving priority to patients with short life expectancy.

Further aims are:

- to understand what factors motivate any observed preference for placing greater weight on a unit of health gain for end of life patients than on that for other types of patients;
- to examine whether the focus on life extensions and absence of quality of life improvements in NICE's end of life policy is consistent with public preferences;
- to assess whether people's stated preferences regarding the value of health gains for end of life patients depend on the ways in which the preference elicitation tasks are designed, framed and presented;
- to contribute to the literature on public preferences regarding the prioritisation of health care.

The scope of the thesis is to present a series of empirical investigations of people's stated preferences regarding the value of health gains for end of life patients. It is acknowledged that there are a number of other potential health care priority-setting criteria and issues that are worthy of investigation. This thesis focuses on just one of these issues, deemed to be topical and highly relevant to current health care policy, though many of the methods and analyses could be applied to the examination of other candidate topics. The scope is also limited to the preferences

of the general public. The preferences of other stakeholder groups, such as patients and policy makers, are not considered.

While the thesis reviews and discusses relevant research undertaken in other countries, all primary empirical research undertaken focuses on the general public in the UK.[1] This reflects the focus on NICE (an agency that operates in England) and the fact that the author resides, works and studies in England.

The weighting of health gains for end of life patients is considered within the framework of cost-effectiveness analysis, a form of economic evaluation that underpins NICE's methods for the assessment and appraisal of health technologies. The remainder of this chapter sets the context for the research in the thesis, providing background information about cost-effectiveness, efficiency and equity objectives of health care, stated preference research, UK policy developments, and the calls for empirical evidence that this thesis has sought to respond to. Brief explanations of the basic concepts used regularly in the thesis are provided in Appendix 1.

1.2 Cost-effectiveness analysis

No health system can afford all health care for everyone, since the resources used to provide health care are scarce. Decisions therefore need to be made about how to allocate health care resources and how to set priorities. Economic evaluation – defined by Drummond *et al.* (2005, p.9) as "the comparative analysis of alternative courses of action in terms of both their costs and consequences" – can be used to guide these decisions. It helps answer the question of whether a particular health technology, intervention or programme is worth investing in, relative to other things that could be done with the same resources. The four main techniques of economic evaluation are cost-minimisation analysis, cost-benefit analysis, cost-effectiveness analysis and cost-utility analysis. Cost-minimisation analysis identifies the least costly option for achieving a fixed outcome. Since it is seldom the case in practice that competing alternatives generate identical outcomes (Brazier *et al.*, 2017), cost-minimisation analysis is rarely used and is not considered further in this thesis.

[1] NICE was initially set up as an England and Wales Special Health Authority, but the way it has been established in legislation means that its guidance is officially England only. However, the Institute has agreements to provide certain services to Northern Ireland, Scotland and (in particular) Wales, and the devolved administrations are often involved and consulted with in the development of NICE guidance. In Wales, there exists a memorandum of understanding which formally sets out the circumstances in which NICE and the All Wales Medicines Strategy Group collaborate in order to provide guidance about treatments funded by NHS Wales (All Wales Medicines Strategy Group, 2016). Hence, this thesis refers at different points to England, England and Wales, and the UK, as appropriate.

Cost-benefit analysis values both the costs and the consequences of the intervention under evaluation in monetary terms. It is widely used in many settings, particularly in the evaluation of transport and environmental programmes (Sugden and Williams, 1979; Mallard and Glaister, 2008; Nas, 2016), and can be described as the broadest form of economic evaluation (Drummond *et al.*, 2005). But cost-benefit analysis is restricted to including only those consequences that can be expressed in terms of money. Well-functioning markets enable goods and services to be valued in monetary terms, reflecting consumers' observed willingness to pay and suppliers' willingness to accept payment for those goods and services. Such 'revealed preferences' are often unavailable in the health setting due to various market failures and other distinctive features, such as information asymmetries and heavily subsidised prices for health care at the point of use. In the absence of readily available information about revealed preferences, 'stated preferences' can be sought via contingent valuation (Donaldson *et al.*, 2006), whereby individuals are asked how much they would be willing to pay for an intervention. However, this too poses challenges as people may not be used to paying directly for health care (in publicly financed systems, for example, the costs of health care are not borne by the beneficiaries but are distributed across the system) or for some of the intangible benefits generated by health care.

For these reasons, cost-effectiveness analysis – which measures consequences in terms of a single natural unit, such as the number of episode-free days – is more commonly used to evaluate health care than cost-benefit analysis. This type of analysis is suitable when comparing interventions that affect a single shared outcome (Weinstein and Stason, 1977). A special type of cost-effectiveness analysis – sometimes referred to as cost-utility analysis (Drummond *et al.*, 2005) uses the quality-adjusted life year (QALY) as that single outcome.[2] The QALY is a generic measure of health that combines quality of life and length of life in a single index (Weinstein *et al.*, 2009). QALYs are calculated by multiplying the number of life years by a numeric quality of life weight. One QALY is equivalent to one year of life in full quality of life (often referred to as full health). Quality of life is measured on a scale that is anchored at one (representing full health) and zero (representing dead). A quality of life weight of less than one implies that the individual in question is not in full health, and a weight below zero implies that the individual is in a state of health considered to be worse than dead. Preference elicitation techniques to obtain quality of life weights for defined health states include time trade-off and standard gamble (Green *et al.*, 2000). Underpinning the QALY model

[2] In this thesis, the term cost-effectiveness analysis is used in favour of cost-utility analysis, in line with much of the literature (Brazier *et al.*, 2017).

is an assumption that the quality of life weight (or value) for a given health state is independent of the duration of that state and when that state occurs (Brazier *et al.*, 2017).

The use of the QALY is contentious (Nord *et al.*, 2009, Neumann, 2011), but its broad and generic nature allows it to act as a 'common currency' for assessing health effects both within and across disease areas. It is therefore widely used by agencies undertaking health economic evaluations – for example, in England (NICE, 2013a), Netherlands (Zorginstituut Nederland, 2015) and Australia (Pharmaceutical Benefits Advisory Committee, 2016). The results of a cost-effectiveness analysis can be expressed in terms of the incremental cost-effectiveness ratio (ICER) of the intervention under evaluation – that is, the incremental cost per QALY gained by investing in the intervention relative to an appropriate comparator. The ICER of that intervention can then be compared to the ICERs of other interventions, or to some threshold value that reflects either displaced activities (that is, the opportunity cost of investing in the intervention) or societal willingness-to-pay for an additional QALY (Towse *et al.*, 2002).

1.3 Health-maximisation

Maximising principles, whereby resources are allocated so as to bring about the best possible consequences, are central to traditional economic analysis. It is widely assumed by health economists that the principal objective of health care is to maximise population health, subject to relevant budget constraints (i.e. what health care resources are available) (Culyer, 1997a; Dolan *et al.*, 2005). Health-maximisation is an attractive objective for those who are concerned about allocating resources efficiently, rather than with regard to equity or social justice (Drummond, 1989). The health-maximisation objective implies a kind of egalitarian approach whereby equal social value is attached to each unit of health gain, regardless of to whom it accrues or the context in which it is enjoyed. This is sometimes referred to as the 'a QALY is a QALY is a QALY' principle (Tsuchiya, 2012).

While clearly linked to the ethical theory of utilitarianism, health-maximisation differs from classical utilitarianism in that the former involves maximising health and the latter involves maximising utility (Culyer, 1989). It is grounded in extra-welfarism – that is, a rejection of the welfarist view that social welfare is simply the sum of individual welfare (utility) of members of the community (Sen, 1979; Culyer, 1989).

Setting priorities using the health-maximisation framework can involve ranking activities such that those "that generate more gains to health for every £ of resources take priority over those that generate less; thus the general standard of health in the community would be correspondingly higher" (Williams, 1985, p.326). Gafni and Birch (1993) describe a 'league table' approach in which activities are ranked in order of their ICERs, and adopting sequentially until the budget is exhausted. However, since this approach involves infeasible informational requirements, in practice other methods are used to define the threshold for determining which activities should and should not be funded. For example, the threshold may be based on an estimate of the opportunity costs of displacing existing activities at the margin in order to fund a new, cost-increasing activity. In that case, the health-maximisation objective dictates that, assuming a fixed budget, the new activity should be funded only if its ICER is lower than that of the activity that would need to be displaced.

Regardless of how the threshold is defined, an outcome of the health-maximisation approach is that when faced with a choice between two equally costly options, the one that is expected to generate greater health gains is the one that will be prioritised, irrespective of any other factors.

1.4 Alternative objectives of health care

While health-maximisation – an example of an efficiency objective – is an important goal in many health care systems, it is unlikely to be the sole goal of either decision-makers or society more generally. Many health policy initiatives in the UK, for example, have been shown not to have been driven by the pursuit of health gains (Shah *et al.*, 2012). It may be legitimate to compromise health-maximisation in order to pursue alternative objectives that allow for equity and other considerations, such as 'improving the health of the poorest fastest' (Department of Health, 2003; Department of Health, 2010a).

Cookson and Dolan (2000) identify maximising principles (maximising aggregate health or aggregate utility) as one of three different classes of principles of justice in health care rationing decisions; the others being egalitarian principles and need principles. The former involves allocating resources so as to reduce inequalities in health. Examples include the 'fair innings' argument for equalising lifetime health (Williams, 1997) and a related argument for equalising people's *opportunities* for achieving lifetime health (Le Grand, 1991). Need principles involve allocating resources in proportion to the degree of need, which can be defined in terms of ill health (immediate ill health, lifetime ill health or threat to life) or in terms of capacity to benefit from treatment. It has also been argued that the primary

objective of the health care system should be to protect the life and health of all individuals, and to give all individuals an equal chance of having their own health needs met (Harris, 1997). This argument has been criticised by health economists for failing to acknowledge the limited availability of resources and for failing to distinguish 'needs' from 'mere wants' (Culyer, 1997b).

Cookson and Dolan (2000) also note that it is possible to combine principles, either by weighting them together or by specifying that a secondary principle is "to come into operation only when the primary principle does not yield a definite answer" (p.327). People may, for example, advocate health-maximisation up to a certain threshold beyond which other principles are considered relevant (Ubel *et al.*, 2000). Egalitarian objectives are typically pursued in combination with other objectives, as blind pursuit of equality is likely to have unreasonable implications for resource allocation.[3]

Based on egalitarian and/or need principles, society may place more importance on a unit of health for some people than on that for other people. If members of society hold preferences as citizens rather than as consumers, motivated by what is beneficial for society, then such preferences can be used to form the basis of policy (Nussbaum, 1999; Dolan *et al.*, 2003; Tsuchiya, 2012).

1.5 Weighting health gains to account for equity considerations

It has been suggested that the best way to combine efficiency and equity concerns is to use societal preferences to construct equity weights to be attached to QALYs (Williams, 1997). Resource allocation decisions could then be based on the incremental cost-per-*equity-adjusted*-QALY gained of competing technologies, with the overall objective of maximising equity-weighted QALYs (Culyer, 1989). Nord (1999), for example, has proposed a system of weighting which involves applying a transformation function in order to give greater weight to gains and losses for those who are more severely ill in terms of quality of life.

However, Wailoo *et al.* (2009) suggest that even if legitimate, valid and reliable weights become available, incorporating these weights into cost-effectiveness analysis remains challenging. A supposedly straightforward approach to incorporating equity weights involves multiplying the number of incremental QALYs generated by a given technology by the relevant equity weight, and comparing the

[3] For example, imagine a situation is which half of the population is healthy and the other half is unhealthy. It seems difficult to justify an egalitarian-driven policy that corrects for this inequality by making both groups unhealthy (albeit equally so).

resulting cost-per-equity-adjusted-QALY to the standard threshold. A mathematically equivalent approach is to use the equity weight to adjust the threshold. Assuming the equity weight is greater than one, this effectively downgrades the QALYs generated by the activity displaced at the margin. As Wailoo *et al.* point out, this means that the QALY gains for patients affected by the displaced activity are being adjusted not according to their own characteristics, but according to the characteristics of a different patient group. But the patients who bear the opportunity costs may themselves have characteristics that warrant special weighting of their health gains – indeed, it cannot safely be assumed that they do not share the same characteristics as the patients who are deemed to be eligible for equity weighting. Leigh and Granby (2016) suggest that equity weights cannot rationally be attached to a technology under evaluation unless it is possible to identify and apply the correct weight to the bearers of the opportunity costs of funding that technology.

A further complication is that, given that many different (and often non-mutually exclusive) equity-related attributes exist, multiple weights would likely apply to the health gains of any given patient group, and these weights may vary over time and act in different directions (Tsuchiya, 2012). A methodologically acceptable functional form for applying multiple weights would need to be identified. The use of weights may also make the resource allocation process more opaque than is desirable. Wailoo *et al.* (2009) warn that while equity weights can facilitate the reallocation of health care resources, they do not increase the total amount of available resources. Equity weighting is therefore an example of a zero sum game: any increases in resources for some patient groups due to unequal weights must be balanced by reductions in resources for other patient groups.

Given the complexity of the various judgements involved, health care decision-makers often prefer to make recommendations on a case-by-case basis[4] via a deliberative process (Culyer, 2009) rather than to rely on an algorithmic weighting scheme. Most jurisdictions that use cost-effectiveness information to guide priority-setting decisions tend to consider equity implicitly, rather than defining a set of explicit weights (Skedgel, 2013). There is increasing interest in understanding how to structure the decision-making process for complex reimbursement decisions involving trade-offs between multiple criteria. A growing number of health care decision-making agencies – including the Institute for Quality and Efficiency in Health Care (IQWiG) in Germany and the Regione Lombardia in Italy – are exploring the application of structured decision-making processes such as multi-

[4] At the technology level, rather than at the individual patient level

criteria decision analysis (Radaelli *et al.*, 2014; Thokala *et al.*, 2016; Devlin and Garau, 2017).

1.6 Role of public preferences

Assuming it is deemed appropriate to apply equity weights to health gains, and irrespective of the precise way in which the weighting system operates, the direction and magnitude of the weights are matters of value judgement (Brazier *et al.*, 2017). A pertinent question to ask is *whose* values should be used.

Since the eventual goal of economic evaluation is to guide policies about resource allocation that fulfil the interests of society as a whole (rather than to make decisions at the individual patient level), it seems appropriate that any social value judgements being fed into the process should be informed by the preferences of a representative sample of society – i.e. members of the general public (Shah, 2009). The importance of public participation in health care decision-making has been emphasised by the government in the UK (Department of Health, 1997), where members of the general public are both potential users and (as taxpayers) the ultimate funders of the country's NHS. Towse (1999) has argued that a compelling reason for public participation in UK health care priority-setting decisions is that since the NHS is a monopoly public service, most people cannot switch to a competing service and so rely on surveys to voice their preferences. The general public may also be expected to be more detached and less biased than other candidate judges (such as clinicians, current patients and unelected health care managers) of the relative value of health gains across different patient groups (Dolan *et al.*, 2003). Empirical studies of public preferences can provide meaningful information as long as the methods used are scientifically defensible (Ryan *et al.*, 2001). Richardson and McKie (2005) have argued that such research should form part of an 'empirical ethics' approach to allocating health care resources.

For these reasons, most empirical studies examining the relative value of a QALY have involved surveys using general public samples (Brazier *et al.*, 2017). A review of public involvement in health care published in 1999 reported that the methods used to understand public views about priority-setting were at that time dominated by opinion polls, satisfaction surveys and citizens' juries (Kneeshaw, 1997). However, economists have argued that in order for public involvement to be useful, the methods used should involve opportunity costs (the notion that providing more of one good means providing less of something else), give an indication of the strength of preference, and involve trade-offs that reflect the kinds of choices that actually have to be made by health care decision-makers (Shackley and Ryan, 1995; Towse, 1999). More recently, public preferences have increasingly been

elicited using 'choice-based methods', which are based on the premise that the value of something can be demonstrated by how much people are willing to sacrifice in order to obtain it. For example, the willingness-to-pay method involves the sacrifice of money, and the person trade-off method involves the sacrifice of benefit to another group. For a review of techniques used to elicit public preferences, see Ryan *et al.* (2001).

It should be noted that not all preferences held by the public are suitable for forming the basis of policy decisions (Tsuchiya, 2012). For example, the public may express a preference for prioritising or restricting health care resources in ways that are legally prohibited or politically unappealing. Nevertheless, public preference studies are useful in that they can help inform decision-makers about the values and priorities of the populations whom they serve.

1.7 Evidence on potential priority-setting criteria

There are a number of published reviews of the empirical evidence on public priority-setting preferences and on the relative social value of a QALY (Schwappach, 2002; Dolan *et al.*, 2005; Whitty *et al.*, 2014; Gu *et al.*, 2015). Researchers have categorised the various factors contributing to the social value of a QALY in different ways. Broadly speaking, these factors may relate to characteristics of patients (such as age and socioeconomic status), characteristics of the health effect (such as the size and direction of the effect), or the way in which the QALYs are distributed (Schwappach, 2002). Other, less frequently studied factors relate to the characteristics of the technology itself – for example, whether it represents a breakthrough innovation that has the potential to promote scientific progress or whether it generates wider societal benefits beyond direct health effects (Linley and Hughes, 2013).

One factor that has been the focus of much debate in the literature is whether greater weight should be placed on QALYs for those who are severely ill. Severity has been the specific focus of a separate review, and is discussed in detail in 1.7.1. Another factor for which there is a relatively rich body of empirical evidence is patient age. The majority of studies indicate that people place greater weight on a unit of health gain for younger individuals than on that for older individuals (Dolan *et al.*, 2005; Gu *et al.*, 2015). However, in some cases preferences regarding age have been found to display a non-linear pattern, with gains for working age individuals valued more than equivalent gains for younger individuals (Whitty *et al.*, 2014; Gu *et al.*, 2015). Gu *et al.* (2015) also warn that preferences for age may be confounded by preferences for a person's capacity to benefit from treatment (in that a younger person has greater to capacity to achieve sizeable life extensions

than does an older person) and that some studies fail to control for this confounding effect. Further, there are some studies that have not supported age as an important factor relative to other potential prioritisation criteria (Whitty *et al.*, 2014).

The evidence base for other factors is less developed, but overall there appears to be a tendency to prefer giving higher priority to: those who are not considered to be responsible for their illness compared to those who are; those with lower socioeconomic status compare to those with higher socioeconomic status; and preventive (health loss-avoiding) interventions over curative (health-improving) treatments.

1.7.1 Severity of illness

This sub-section focuses on severity of illness as a priority-setting or QALY weighting criterion. Severity is particularly relevant to this thesis due to its clear overlap with end of life – indeed, the latter may be interpreted as a subset of the former.

A concern for the worst off in society is a common feature of several different principles of justice (Rawls, 1972; Daniels, 1985; Brock, 2001), including both egalitarian and need principles. While the definition of need in terms of capacity to benefit may lead to the objective of QALY-maximisation, other definitions of need – based on immediate ill health, lifetime ill health, or threat to life – lead to a competing objective of placing special weight on the QALYs accruing to those who are severely ill (described by Nord (2005) as the 'severity approach'). This is also consistent with a basic principle governing the actions of clinicians, who generally seek to prioritise according to the perceived degree of suffering (Cubborn, 1991).

Severity can be defined in different ways. A person can be described as severely ill if their expected number of lifetime QALYs represents a major shortfall (in either absolute or proportional terms) from a 'normal' QALY expectancy (Williams, 1997; Towse and Barnsley, 2013). Alternatively, a person can be described as severely ill if their current level of health is poor. The severity approach suggests that the value of a health gain would depend on the patient's level of (lifetime or current) health prior to benefiting from that gain.

A review of the empirical evidence on severity-related preferences published in 2009 suggests that people are, on the whole, willing to sacrifice aggregate health in order to give priority to the severely ill (Shah, 2009) – 17 of the 19 studies

reviewed reported results broadly consistent with the severity approach.[5] Similar findings have been reported in more recent reviews (Nord and Johansen, 2014; Whitty *et al.*, 2014; Gu *et al.*, 2015). The majority of studies reviewed by Shah (2009) focused on severity as measured by a patient's current quality of life in absence of treatment, with several using adapted versions of a simple mobility scale used by Nord (1993). Few studies defined severity in terms of length of life or proximity to death. In some cases, life expectancy was controlled for by asking respondents to assume that it did not differ across patients. In other cases, it was less clear how life expectancy (and duration more generally) was to be considered.

The importance of severity is already well established in a number of jurisdictions (Shah, 2009). Since the 1980s, the Norwegian National Health Service has recognised that severity should be considered alongside the effectiveness of treatment when prioritising between patients (Norwegian Commission for Prioritising in Healthcare, 1987). A recent Norwegian government commission on priority-setting recommended that for group-level decisions, severity should be measured in terms of the absolute loss of QALYs as a result of not making available the treatment under assessment (Ottersen *et al.*, 2016). This recommendation has just been approved and endorsed by the Norwegian government (Norwegian Ministry of Health and Care Services, 2017). In Australia, the Pharmaceutical Benefits Advisory Committee (2016) applies a so-called 'rule of rescue' whereby special consideration may be given to a treatment for a condition that meets the four factors concurrently, one of which being that the condition is severe.[6] In the Netherlands, an equity principle called 'proportional shortfall' – a measure of severity based on the proportion of lifetime health lost due to illness – has been proposed (Stolk *et al.*, 2004). According to van de Wetering *et al.* (2013), there has been support for incorporating information about proportional shortfall into Dutch technology assessments. However, its influence in decision-making has been limited by a lack of consensus on how best to operationalise a QALY weighting system based on proportional shortfall and on whether such a system would be aligned with societal preferences (van Exel, J., 2017, personal communication, 16 February). In Sweden, the Dental and Pharmaceutical Benefits Agency assesses technologies in relation to a threshold that reflects individuals' maximum

[5] Shah (2009) defined the severity approach as follows: "when conducting CUA [cost-utility analysis] in order to make allocation decisions, the health gains accruing to individuals in poorer health without treatment should be valued more highly than those accruing to individuals in better health without treatment, with health defined in terms of quality of life, length of life, or both" (p.79).

[6] Note that the Australian operationalisation of the rule of rescue does not focus on the rescue of identifiable individuals from imminent death, and is therefore substantially different from the original rule of rescue discussed in the literature (Cookson *et al.*, 2008). McKie and Richardson (2003) argue that it is the emphasis on identifiable individuals that distinguishes the rule of rescue from a more general concern for the worst off underpinning the severity approach.

willingness-to-pay for a QALY. The threshold is adjusted for need, which is related directly to disease severity (defined as the expected loss of QALYs for untreated patients, though a strict formula for determining the degree of severity is not applied) (Persson, 2012).

1.8 UK policy context

1.8.1 NICE and its social value judgements

NICE provides national guidance and advice on the use of health technologies in the NHS in England (and in certain cases in Northern Ireland, Scotland and Wales). It was set up in 1999 as a special health authority (that is, an arm's length body of the Department of Health), primarily to ensure that everyone living in England and Wales had equal access to health care from the NHS. Since April 2013 it has been established as a non-departmental public body. It is accountable to, but operationally independent of, the UK government.

NICE's activities include a technology appraisal programme (which includes, *inter alia*, the assessment and appraisal of branded medicines), an interventional procedures programme, a highly specialised technologies programme, a medical technologies evaluation programme (covering devices and diagnostics) and the development of clinical guidelines (Cowles *et al.*, 2017). NICE also produces public health and social care guidelines.

NICE's technology appraisals are guided by clinical and cost-effectiveness analyses, usually using the QALY to measure health outcomes (and with the EQ-5D specified as the preferred measure of the quality of life component) (NICE, 2013a). The general requirement is that in order to be recommended for routine use in the NHS, the technology under appraisal should have an ICER at or below a threshold range of £20,000 to £30,000 per QALY gained. The likelihood of a given technology being rejected on grounds of cost-ineffectiveness increases as its ICER increases. A technology with an ICER below £20,000 per QALY gained is likely to be recommended, while the acceptability of a technology with an ICER of between £20,000 and £30,000 per QALY gained is more likely to depend on other factors, such as the innovative nature of the technology and aspects that relate to non-health objectives of the NHS. An increasingly strong case with regard to these factors would need to be made in order to recommend the use of a technology with an ICER above £30,000 per QALY gained (NICE, 2013a).

Current guidelines used by NICE define a 'reference case' position. This position corresponds to the aforementioned 'a QALY is a QALY is a QALY' principle: "In the

reference case, an additional QALY should receive the same weight regardless of any other characteristics of the people receiving the health benefit" (NICE, 2013a, paragraph 5.4.1). In the first edition of its technology appraisal methods guide, NICE explained that this position "reflects the absence of consensus regarding whether these or other characteristics of individuals should result in differential weights being attached to QALYs gained" (NICE, 2003, paragraph 5.9.7.2).

However, NICE's technology appraisal committees (independent advisory committees responsible for formulating guidance) are given discretion to consider alternative equity positions and are expected to make social value judgements. These are concerned with what is appropriate and acceptable for society in delivering health care across the NHS. The appraisal committees occasionally depart from 'a QALY is a QALY is a QALY' principle in order to incorporate values reflected in legislation, in the deliberations of NICE's Citizens' Council (see below), and in feedback from stakeholders such as patient representatives and health care professionals (Chalkidou, 2012).

NICE is unusual amongst similar organisations elsewhere in the world in that it systematically publishes details of the evidence and reasoning underpinning its decisions, including references to social value judgements (Shah *et al.*, 2013a). However, while its principles related to cost-effectiveness are relatively explicit (NICE, 2013a), those covering equity concerns have generally been less specific. NICE's approach to social values is not driven by a single philosophical perspective but is pluralistic in that it draws on several traditions (Shah *et al.*, 2013a).

NICE and its advisory committee are well placed to make scientific value judgements but have no special legitimacy to impose social value judgements on the NHS. The Institute's position is that "advice from NICE to the NHS should embody values that are generally held by the population that the NHS serves" (Rawlins and Culyer, 2004, p.226). For this reason – and in line with the NHS's increasing emphasis on the need for public involvement in health care decision-making (Department of Health, 1997) – NICE formed a Citizens' Council in 2002. The Council is a panel of 30 members of the public that is intended to broadly reflect the demographic characteristics of the general population (NICE, 2017a). It normally meets once a year to discuss and provide NICE with a public perspective on matters of ethics and equity that have arisen during guidance development activities. Topics discussed by the Council include the definition of clinical need (NICE, 2002a), the legitimacy of adopting a 'rule of rescue' criterion (NICE, 2006), the use of severity as a priority-setting criterion (NICE, 2008a) and the appropriate reasons for deviating from the usual threshold range (NICE, 2008b). The Council's

recommendations – which are reached following a structured deliberative process (Culyer and Lomas, 2006) – are incorporated into a general guidance document called *Social value judgements: Principles for the development of NICE guidance* (NICE, 2008c) and, where appropriate, into the technology appraisal methods guide.

Accordingly, NICE's appraisal committees have occasionally applied social value judgements and 'special weighting' in order to recommend treatments with ICERs exceeding the range normally considered acceptable (Rawlins *et al.*, 2010; Shah *et al.*, 2013a). For example, in the appraisal of riluzole for motor neurone disease, the committee considered the 'severity and relatively short lifespan' of affected patients, and recommended the use of the technology despite the fact that the ICER estimates ranged between £34,000 and £43,500 per QALY gained (NICE, 2001). Similarly, life-extending trastuzumab combination therapy was recommended for use in the treatment of advanced breast cancer even though the manufacturer provided an ICER estimate of £37,500 per QALY gained. The decision was due in part to the committee's observation that "improvements in survival of this magnitude due to therapeutic intervention have rarely been recorded in women with metastatic breast cancer" (NICE, 2002b, paragraph 4.3.3). According to Rawlins *et al.* (2010), NICE's appraisal committees have also sought to take into account whether the treatments under appraisal target children and/or disadvantaged populations. However, until 2009 there were no official circumstances in which appraisal committees were asked to depart *systematically* from the reference case position.

1.8.2 NICE's guidance on appraising life-extending end of life treatments

In 2008, several high-profile rejections of new treatments for renal cancer on cost-effectiveness grounds resulted in criticism from the media, patients and pharmaceutical companies (Wagstaff, 2008; Walker *et al.*, 2008), and in calls for a more generous coverage policy for cancer treatments. Some patients sought to supplement their free NHS care by paying for these treatments out-of-pocket, but the government ruled that such 'top-ups' were not permitted (Timmins *et al.*, 2016). Under mounting pressures, the Department of Health announced a four-month review to examine "if, when and in what circumstances patients should be able to purchase additional drugs that are not funded by the NHS" (Richards, 2008, p.2). The review recommended that, *inter alia*, NICE should demonstrate greater flexibility and assess "what measures could be taken to make available drugs used

near the end of life that do not meet the cost-effectiveness criteria applied to all drugs" (p.4).

The government accepted the recommendations and called on NICE to review its methods for appraising end of life treatments (Chalkidou, 2012). NICE began a public consultation on proposed supplementary advice for its appraisal committees. It was originally proposed that appraisal committees should consider recommending the use of medicines with an ICER exceeding the range normally considered acceptable if *all* of the following criteria were met (NICE, 2009a):

- The medicine is indicated for the treatment of patients with a diagnosis of a terminal illness and who are not, on average, expected to live for more than 24 months.
- There is sufficient evidence to indicate that the medicine offers a substantial extension to life, compared to current NHS treatment.
- The medicine is indicated, in its licence, for a patient population normally not exceeding 7,000 new patients per annum.

Some aspects of the supplementary advice were revised following the consultation exercise (NICE, 2009b). Notably, the 'substantial extension to life' criterion was replaced by a more specific threshold of a three-month extension, and the 7,000 patient threshold was replaced by a less specific 'small patient populations' criterion. Further, an earlier proposal – that in order for a treatment to be eligible for special consideration there should be a lack of alternative treatments with comparable benefits available in the NHS – was dropped (Chalkidou, 2012). The revised criteria (NICE, 2009c) are reproduced below; if met, the appraisal committees were asked to consider the impact of giving greater weight to the health gains achieved in the later stages of disease:

- The treatment is indicated for patients with a short life expectancy, normally less than 24 months.
- There is sufficient evidence to indicate that the treatment offers an extension to life, normally of at least an additional three months, compared to current treatment.
- The treatment is licensed, or otherwise indicated, for small patient populations.

The advice stated that the appraisal committees should be satisfied by the robustness of the estimates and assumptions used in the economic modelling and that all calculations should consider the cumulative population for each licensed indication. It also asked the committees to consider:

- The impact of giving greater weight to QALYs achieved in the later stages of terminal diseases, using the assumption that the extended survival period is experienced at the full quality of life anticipated for a healthy individual of the same age, and;
- The magnitude of the additional weight that would need to be assigned to the QALY benefits in this patient group for the cost-effectiveness of the technology to fall within the current threshold range.

No reason was specified for why it should be assumed that the life extension would be experienced at full quality of life. The consultation response document (NICE, 2009b) referred to a number of arguments made in favour of placing greater weight on life extensions for end of life patients. These included: the argument that such a policy was consistent with societal values; the aspiration for access to end of life drugs to be 'in step with' the levels observed in other high-income countries; the importance of supporting the development of innovative treatments; and the concern that the standard QALY model fails to adequately capture important benefits for end of life patients. The latter argument is discussed in detail elsewhere (Garau *et al.*, 2011; Round, 2012; Devlin and Lorgelly, 2017). The supplementary end of life policy represented the first systematic departure from the 'a QALY is a QALY is a QALY' reference case position underpinning NICE's technology appraisals. It has been referred to in the literature as an 'end of life premium' (Cookson, 2013; Linley and Hughes, 2013; McCabe *et al.*, 2016a).

Of the first 23 technologies that were considered in light of the supplementary advice, 13 were considered to have met all of the criteria. Of those 13, eight were ultimately recommended for use in the NHS (Trowman *et al.*, 2011). More recent analysis by Barham (2016) suggests that the effective threshold applied to end of life treatments between 2009 and 2015 was around £49,000 per QALY gained. Other sources refer to an unofficial threshold of £50,000 per QALY gained for end of life treatments (NICE, 2013b; Boysen, 2014; Stewart *et al.*, 2014; Parliamentary Office of Science and Technology, 2015).

1.8.3 Selected criticisms of NICE's end of life policy

While NICE's end of life policy has resulted in increased access to end of life treatments for patients, if the NHS budget is fixed then the funds for these treatments must be found by disinvesting from other activities. Collins and Latimer (2013) have shown, under the assumption that society places no more weight on a unit of health gain for end of life patients than on that for other types of patients, that the application of NICE's policy "is likely to have resulted in substantial QALY losses and budgetary pressures to the NHS and population in England and Wales,

as cost effective interventions are displaced in favour of less cost effective interventions" (p.2).[7] However, the authors acknowledge that if society does in fact place greater weight on health gains at the end of life, then the overall QALY loss may be acceptable to the extent that it represents societal preferences.

In a paper addressing the normative issues raised by NICE's policy, Cookson (2013) considers whether 11 potentially relevant ethical arguments can be used to provide support for an end of life premium. Seven of the arguments[8] are rejected on the basis that they are not relevant in the context of a national agency making recommendations about the public funding of health technologies based on the systematic assessment of value for money. The other four arguments[9] are rejected on the basis that "applying them systematically would yield something quite different from the NICE end-of-life premium" (p.1145). These include the argument that health care resources should be allocated in relation to severity – Cookson points out that proximity to death is an excessively narrow way to define severity, and contrasts the binary nature of NICE's end of life cut-offs to the continuous nature of typical definitions of severity.

Paulden *et al.* (2014) similarly highlight the arbitrary nature of the cut-offs in the criteria – for example, a treatment for patients with *just under* two years of life expectancy in *good* quality of life would meet the end of life criteria whereas a treatment for patients with *just over* two years of expectancy in *poor* quality of life would not. The authors point out the inconsistency of quality of life improvements being central in NICE's assessments of the cost-effectiveness of health technologies, yet irrelevant to its end of life criteria.

Raftery (2009) has warned that making an exception to the usual rules in order to recommend the use of life-extending end of life treatments limits the universality of the cost-per-QALY approach and sets a precedent that could lead to special pleading by other groups.

1.8.4 Recent policy developments

This sub-section provides details of policy developments in England that followed the introduction of NICE's end of life guidance in 2009, including the planned value-based pricing and value-based assessment schemes and the introduction of a ring-

[7] Specifically, Collins and Latimer (2013) estimate a net annual loss of between 5,933 and 15,098 QALYs depending on which value of the standard cost-effectiveness threshold is assumed.
[8] Rule of rescue; fair chances; *ex post* willingness-to-pay; caring externality; financial protection; symbolic value; diminishing marginal value of future life years. Cookson acknowledges that some of these arguments may be relevant considerations in other decision-making contexts, such as how politicians or health care professionals should respond to exceptional, emergency situations.
[9] Concentration of benefit; dread; time to set your affairs in order; severity of illness.

fenced 'Cancer Drugs Fund' (CDF). Although these policies are not the focus of this thesis, they are relevant because they represent further departures from the 'a QALY is a QALY is a QALY' position, and (could have) had implications for the appraisal of and access to end of life treatments.

In 2010, the Department of Health published a consultation document on a proposed scheme for the value-based pricing of branded medicines. One of the stated objectives of the scheme was to "include a wide assessment, alongside clinical effectiveness, of the range of factors through which medicines deliver benefits for patients and society" (Department of Health, 2010b, paragraph 3.3). This indicated that the 'value' of a medicine is derived not only from the health gains from treatment, but also from other factors that may be of benefit to society. Under the new system, it was proposed that in addition to the basic threshold reflecting benefits displaced in order to fund new medicines, there would also be higher thresholds for medicines for diseases associated with high levels of 'burden of illness': "the more the medicine is focused on diseases with unmet need or which are particularly severe, the higher the threshold" (paragraph 4.10). The consultation document did not state how severity and unmet need were to be defined. Higher thresholds were also proposed for medicines that demonstrated "greater therapeutic innovation and improvements compared with other products" and/or "wider societal benefits" (paragraph 4.10).

A Health Select Committee report into NICE, published in 2013, criticised the delays in developing and implementing the value-based pricing scheme (Health Committee, 2013). In response, the Department of Health asked NICE to take into account additional terms of reference for the 'value based assessment of health technologies' (NICE, 2013b). This requested that NICE's methods should, *inter alia*, "include a simple system of weighting for burden of illness that appropriately reflects the differential value for the most serious conditions" (p.15). It was suggested that burden of illness could be estimated "using a simple percentage weighting that is proportionate to the QALY loss suffered by patients with the condition" (p.15). Accordingly, NICE developed a set of proposals to incorporate these new terms of reference into its appraisal methods. These included the proposal that burden of illness would be assessed by calculating patients' absolute and proportional QALY shortfall from normal healthy life expectancy,[10] and that this

[10] Absolute and proportional QALY shortfall can be described as two possible operationalisations of severity of illness (see 1.7.1). According to Towse and Barnsley (2013), absolute shortfall is given by subtracting current health prospects (with condition) from total potential health from today (without condition); and proportional shortfall is given by dividing absolute shortfall by total potential health from today (without condition). These can be contrasted to the fair innings approach, which considers one's QALY shortfall *from birth* rather than from today. The levels of absolute and proportional QALY shortfall

– in combination with other modifiers – would form the basis for QALY weights. It was also proposed that the end of life criteria would be 'subsumed' within burden of illness.

The proposals were outlined in a consultation paper, responses to which were received from patient groups, clinicians, academics, the pharmaceutical industry and other interested groups. No consensus emerged, with respondents particularly split in their views about how burden of illness and other criteria should be measured and valued. As a result, NICE recommended to its Board that no changes to the technology appraisal methodology be made in the short term (NICE, 2014a). This meant that the supplementary end of life policy was retained.

The CDF was introduced in 2010, with the aim of improving access to cancer medicines that were not routinely available in the NHS. It made available cancer drugs that had been rejected by NICE on cost-effectiveness grounds, had not yet been appraised by NICE, or were being used outside of their marketing authorisations. The CDF was intended only as a temporary arrangement prior to the renegotiation of the Pharmaceutical Price Regulation Scheme and the introduction of the aforementioned value-based pricing scheme (Dixon *et al.*, 2016).

According to impact assessment of the CDF proposal, the purpose of the fund was to "enable cancer treatments to be funded by the NHS where society values their benefits more than the benefits that could be provided by spending the funding on other treatments, elsewhere in the NHS" (Department of Health, 2010c, p.1). If society indeed places special value on the health gains achieved by cancer patients, then "cancer treatments which provide less health benefits than the alternative use of funds might still be socially more valuable than the alternatives" (p.12). The extent to which this is the case has been examined elsewhere (Linley and Hughes, 2013; Culyer, 2017; Shah, 2017).

The CDF has been criticised by health economists, who have argued that it undermines the function of NICE, reduces incentives for pharmaceutical manufacturers to lower prices, and arbitrarily singles out cancer for special consideration for emotive reasons (Maynard and Bloor, 2011; Appleby, 2014; Buxton *et al.*, 2014; Claxton, 2015). More generally, it has been argued that the opportunity cost of prioritising cancer drugs tends to fall on lower profile areas of health care (Adams, 2011) such as mental health and palliative care (Barrett *et al.*, 2006).

increase for life-limiting illnesses, particularly when patients are expected to fall short of a 'normal' life expectancy.

A reconfiguration of the CDF was announced in 2016. Under the new system, all new cancer drugs (as well as those already funded via the CDF) are to be referred to NICE's technology appraisal process. For drugs that display plausible potential for satisfying the criteria for routine use (with or without the application of the end of life criteria), a conditional recommendation can be made. In such cases, the drugs would be funded via a new CDF for an agreed period whilst further clinical and cost-effectiveness evidence was collected, with the view to recommending the drugs for routine use subject to the approval of the new evidence (Dixon *et al.*, 2016). Recent analysis suggests that the vast majority of the product-indication pairs that were included in the original CDF would not have met NICE's criteria for defining a life-extending end of life treatment, and most are unlikely to be funded by the revised CDF (Britton, 2016). While the revised CDF addresses some of the problems associated with its predecessor, the notion of a dedicated fund and special access mechanism for cancer drugs remains controversial (McCabe *et al.*, 2016b).

Linked to the new CDF, the NICE end of life criteria were revised in July 2016 as an addendum to the Institute's technology appraisal methods and processes guides (NICE, 2016). The small patient population criterion has been dropped altogether. The current guidance is shown in Box 1 [formatting in original].

At the time of writing, the revised CDF is in place, while NICE is continuing to investigate the possibility of incorporating a system of burden of illness weighting into its methodology (Boysen, 2014). The author's understanding is that the end of life policy is outside the remit of these investigations.

Box 1. NICE's supplementary advice for the appraisal of life-extending end of life treatments (latest version)

6.2.10 In the case of a 'life-extending treatment at the end of life', the Appraisal Committee will satisfy itself that all of the following criteria have been met:

- the treatment is indicated for patients with a short life expectancy, normally less than 24 months and
- there is sufficient evidence to indicate that the treatment has the prospect of offering an extension to life, **normally** of a mean value of at least an additional 3 months, compared with current NHS treatment.

~~and~~

- ~~the technology is licensed or otherwise indicated, for small patient populations normally not exceeding a cumulative total of 7000 for all licensed indications in England.~~

In addition, the Appraisal Committees will need to be satisfied that:

- the estimates of the extension to life are *sufficiently* robust and can be shown or reasonably inferred from either progression-free survival or overall survival (taking account of trials in which crossover has occurred and been accounted for in the effectiveness review) and
- the assumptions used in the reference case economic modelling are plausible, objective and robust.

6.2.11 When the conditions described in section 6.2.10 are met, the Appraisal Committee will consider:

- the impact of giving greater weight to QALYs achieved in the later stages of terminal diseases, using the assumption that the extended survival period is experienced at the full quality of life anticipated for a healthy individual of the same age and
- the magnitude of the additional weight that would need to be assigned to the QALY benefits in this patient group for the cost effectiveness of the technology to fall within the normal range of maximum acceptable ICERs, with a maximum weight of 1.7.

Source: NICE (2016)

1.9 Calls for empirical evidence of societal preferences regarding end of life treatments

It has been claimed that NICE's end of life policy reflects a recognition that society places special value on treatments that extend the life of patients with terminal illness, as long as the life extension is of a reasonable quality (Rawlins *et al.*, 2010). However, the consultation on NICE's end of life policy revealed concerns that there is little evidence to support the premise that society is prepared to fund life-extending end of life treatments that would not meet the cost-effectiveness criteria used for other treatments (NICE, 2009b). The Citizens' Council had expressed an overall preference for taking disease severity into account (NICE, 2008a) and for relaxing the cost-effectiveness threshold under certain circumstances (including for life-saving interventions) (NICE, 2008b). However, the Council had not been asked for its views on the specific criteria underpinning the supplementary policy. Further, a general limitation of the Council is that as a panel of just 30 people, it could only offer limited insight into the views of wider society.

As a result, a number of calls for research on society's preferences were made. A review undertaken by the Department of Health (2010c) to assess the potential impact of the CDF noted the need for robust evidence to support the weighting of health gains accruing to patients who are severely ill or at the end of life. NICE itself acknowledged the need for further testing of the assumptions behind the end of life policy (Longson and Littlejohns, 2009). In an unpublished review of the empirical ethics literature, Green (2011) noted that the evidence on end of life-related preferences was limited and called for further exploration of the issues. More recently, two former chairs of the Scottish Medicines Consortium claimed (referring to the premia for end of life treatments implied by NICE's end of life policy, the CDF, and Scotland's New Medicine Fund) that there "has been tacit acceptance that the changes made match the views of UK society, yet there has been no exploration of whether that is, in fact, the case" (Webb and Paterson, 2016, p.2).

1.10 Structure of the thesis

The remainder of the thesis is divided into six chapters.

Chapter 2 reviews the published literature that is relevant to the overall research question underpinning the thesis. It identifies the extent to which public preferences on this topic have been studied and reported in peer-reviewed journals. It also reports the methods used to elicit preferences and the findings of the existing studies. The search for literature was first carried out in 2014, and

repeated in 2016 in order to update the review. The timing of the review was such that it includes articles based on the studies reported in Chapters 4 and 5.

Chapter 3 describes the first of four empirical studies undertaken – an exploratory study conducted in early 2011 using a convenience sample of 21 members of staff and students at the University of Sheffield. It develops and pilots an approach to examining end of life-related preferences using a choice exercise administered in face-to-face interviews.

Chapter 4 describes the second of the four empirical studies – a study conducted in mid-2011 using a sample of 50 members of the public. This study uses a similar approach to that used in the previous study, though the survey design was refined in accordance with the earlier findings. Five hypotheses relating to the value of life-extending end of life treatments are tested.

Chapter 5 describes the third of the four empirical studies – a web-based discrete choice experiment (DCE) conducted in 2012. The study uses a sample of 3,969 members of the public, broadly representative of the general population in terms of age, gender and social grade. The study design presents respondents with choices representing trade-offs between achieving larger QALY gains and prioritising the treatment of patients with shorter life expectancy. The extent to which the cut-offs implied by the NICE end of life criteria are consistent with public preferences is analysed and discussed in this chapter.

Chapter 6 describes the fourth and final empirical study – a web-based study conducted in 2016 using a sample of 2,401 members of the public, representative of the general population in terms of age, gender and social grade. As well as seeking to address the overall research question underpinning the thesis, this study also seeks to examine the extent to which people's stated preferences regarding end of life treatments are sensitive to a number of framing effects and study design choices. Nine hypotheses are tested, all of which arose from trends, ambiguities and gaps in the existing evidence, as identified in the previous chapters.

Chapter 7 concludes by drawing together and discussing the findings of the overall thesis. It summarises the main contributions and limitations of the research, and identifies some policy implications of the findings and recommendations for further research.

2 DO 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? A REVIEW OF THE LITERATURE[11]

2.1 Introduction

The aim of this chapter is to review the published social sciences literature that is relevant to the following research question: do 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? Policies reflecting such preferences can be described as an 'end of life premium' (see 1.8.2). The need for an up-to-date review is clear – most existing reviews of the empirical ethics literature did not include end of life as an attribute of interest, and two that did are limited in that they are unpublished, did not use systematic methods, and are relatively old (Green, 2011; Keetharuth *et al.*, 2011).

The review reported in this chapter seeks to identify the extent to which public preferences on this topic have been studied in the peer-reviewed literature. It also seeks to provide an in-depth account of the methods used to elicit preferences and the findings of the studies, with the intention of informing policy decisions and future research, including research for this thesis. The decision to focus on the social sciences literature was made based on an informal scoping exercise involving preliminary searches of PubMed Central. These searches indicated that the biomedical literature contained a very large number of articles that would be identified using search terms such as *end of life* but would not be informative to the research question.

The review focuses on studies concerned with the prioritisation of treatment based on patients' life expectancy (or proximity to death), thus distinguishing it from previous reviews of severity of illness more generally (Shah, 2009; Nord and Johansen, 2014), which typically have examined studies describing severity in terms of quality of life.

The review has also applied a creative approach to searching the literature for articles that are relevant to a particular health policy issue.

[11] Selected findings from this chapter have been published as: Shah, K.K., 2016. Does society place special value on end of life treatments? In: Round, J., ed. *Care at the end of life: an economic perspective*. Cham: Springer.

2.2 Methods

2.2.1 SSCI search

The primary source of data for the review was an electronic search of the Social Sciences Citation Index (SSCI) within the Web of Science Core Collection, first carried out in May 2014. The search was repeated in May 2016 in order to update the review. No time or language limits were imposed, though the database only covers articles published since 1956.

An iterative approach was used to identify search terms. The following sub-section therefore includes selected intermediate results, as necessary to explain the methods.

2.2.1.1 Search terms

Two types of search were considered: title term and topic term. The former searches only for terms in the titles of articles. The latter searches for terms in the titles and abstracts of articles, as well as in the keywords that have been assigned to the articles by the authors and Web of Science. The search was based on topic terms for two reasons. The first is that some authors use titles that do not reflect the content of their articles. The second is that some authors may have examined preferences regarding end of life treatments as one of several elements in their study, in which case a term such as *end of life* would be likely to appear in the abstract but unlikely to appear in the title. A full text search was not used due to an expectation that this would reduce the specificity of the search to an unacceptable degree.

Search terms were developed using an iterative process. The initial search terms were *end of life* and *preferences*.[12] In order to improve the sensitivity of the search, two terms related to end of life – *severity* and *terminal* – were added. In a review of severity as a priority-setting criterion (Shah, 2009), some of the studies identified measured severity in terms of life expectancy. In its supplementary guidance on the appraisal of life-extending end of life treatments, NICE (2009c) refers to the benefits of such treatments being achieved "in the later stages of terminal disease" (paragraph 2.2.1).

A form of 'word frequency analysis' (Glanville *et al.*, 2006) was then used to identify further search terms, in order to improve the specificity (and therefore the efficiency) of the search. Three articles were designated as 'key papers' (Abel

[12] Note that Web of Science automatically helps to find plurals and variant spellings, so it makes no difference whether the search term is, say, *preference* or *preferences*.

Olsen, 2013; Linley and Hughes, 2013; Shah *et al.*, 2014) and their abstracts were examined. These were the only three fully-published articles that explicitly investigated public preferences regarding end of life treatments, as defined by the NICE guidance (and therefore of direct relevance to the policy issue that motivated this programme of research), that were known to the author at the time of developing the search strategy. Some discussion/working papers that addressed the topic were also known to the author, but these articles were not used for the purpose of identifying search terms because they had not yet been published in peer-reviewed journals and their abstracts were therefore subject to change.

All of the unique words that appeared in at least two of the three key paper abstracts were identified, and those considered to be potentially relevant to the research question (see 2.3.1 for more details) were selected. The impact of adding these terms to the search was tested by examining whether their inclusion substantially reduced the number of records identified whilst increasing the specificity of the search. This was judged informally by assessing the number of 'probably relevant' records within the most recent 20 records. As a result of this process, the terms *health* and *respondents* (or its synonyms: *subjects*, *participants*, *sample*) were added. The term *life expectancy* was also added as a further alternative to *end of life*. The informal assessment process revealed that at least some of the 56 additional records identified by adding *life expectancy* to the search strategy were potentially relevant, thus justifying the inclusion of the term.

Adding combinations of the terms *patient*, *treatment*, *evidence*, *public* and *population* (as well as related terms such as *popular*) further reduced the number of records identified. However, it was noted that some potentially relevant records were missed as a result of adding these terms. The terms were therefore not included in the search strategy.

Box 2 shows the final strategy (note that TS refers to topic search). The results of preliminary searches are presented in Appendix 2.

Box 2. Final search strategy

```
    TS=("end of life" OR severity OR terminal OR "life expectancy")
AND TS=(preferences)
AND TS=health
AND TS=(respondents OR subjects OR participants OR sampl*)
```

The chosen strategy required that the term *preferences* was included as a topic term. It did not include alternatives to *preferences*, hence it cannot be claimed that a fully exhaustive search has been conducted. Such terms were identified by

examining the titles of empirical studies included in a similar review (Shah, 2009): *attitudes* (as in Oddsson, 2003), *choices* (e.g. as in Ubel *et al.*, 1996), *utilities* (e.g. as in Ubel *et al.*, 1996) and *values* (e.g. as in Dolan and Green, 1998). Including these four terms as alternatives to *preferences* increased the number of results substantially (nearly five-fold). However, an informal assessment of the titles of (a randomly selected sample of) 50 of the additional records identified by adding these terms did not detect any that were considered relevant to the research question.

2.2.2 Selection of studies for inclusion

To be included, articles had to meet all of the following hierarchical criteria:

1. **Publication**: Article must be published in English in a peer-reviewed journal.
2. **Empirical data**: Article must review, present or analyse empirical data.
3. **Priority-setting context**: Article must relate to a health care priority-setting or resource allocation context. Articles reporting preferences from an individual or 'own health' perspective (rather than a social decision-maker perspective) can be included as long as they clearly seek to inform health care priority-setting policies.
4. **Stated preference data**: Article must report preferences that were elicited in a hypothetical, stated context using a choice-based approach involving trade-offs.
5. **End of life**: Article must inform the topic of placing greater weight on a unit of health gain for end of life patients (i.e. patients with short life expectancy) than on that for other types of patients.
6. **Original research**: Article must present original research and must not be solely a review of the literature.

Criterion 3 was applied to ensure that the review focused on studies that can inform the kinds of priority-setting policy issues faced by NICE and other similar agencies. The exclusion of articles reporting preferences only from an individual or 'own health' perspective was considered, as the legitimacy of using such studies to inform decisions about how to allocate shared resources has been questioned (Brouwer and Koopmanschap, 2000). However, it was deemed appropriate not to apply this exclusion rule on the basis that the own health perspective studies may provide information that is relevant to the research question.

Titles and abstracts were screened for eligibility against criteria 1 to 4, sequentially. The full texts of potentially eligible articles were then screened against criteria 1 to 6, sequentially. Full texts were also screened in cases where it was not clear from the title and abstract which of the criteria had and had not been met.

Whitty *et al.* (2014) and Gu *et al.* (2015) both note that there is currently no single standardised method for assessing the quality of stated preference studies covering the full range of preference elicitation techniques (though best practice guidelines do exist for specific methods – e.g. Bridges *et al.*, 2011). Hence, a formal assessment of study quality was not undertaken.

2.2.3 Identification of additional material

Additional material was identified by following up the reference lists of the articles whose full texts were screened. The same criteria were applied to determine whether or not to include these newly identified articles.

2.2.4 Data extraction

Data were extracted for each included study and compiled in an Excel database. Table 2-1 shows the fields used to categorise and describe the studies. Following Whitty *et al.* (2014), it was deemed inappropriate to synthesise the preference data due to the variation in methods and contexts between studies, so a largely descriptive reporting approach was used.

The 'other factors examined' field covers attributes that varied across the tasks, options or choices in a given study, in addition to the attribute(s) of direct relevance to end of life. Information on attributes that were mentioned but held fixed in all tasks, or on factors that were addressed in the study but not in the tasks relevant to end of life, was not extracted systematically.

The last two fields, which concern age- and time-related preferences, were included as these are potential drivers of decisions to prioritise the treatment of end of life patients. When asked whether to give life-extending treatment to patients with shorter life expectancy or to patients with longer life expectancy, there may be reasons for favouring the former other than a preference for treating end of life patients *per se*. First, if all of the potential recipients are the same age today, then in absence of treatment the patients with shorter life expectancy will be younger when they die than the patients with longer life expectancy will be when they die. Hence, a preference for treating patients with shorter life expectancy may be driven by a preference for treating the young. Second, the benefit from treating the patients with longer life expectancy would not take place until further into the

future. Hence, a preference for treating patients with shorter life expectancy may be driven by respondents' social time preference, which is often assumed to be positive. It is therefore informative to examine whether studies attempted to control for or to analyse the impact of age- and time-related preferences.

Table 2-1. Data extraction fields and descriptions

Field	Options (where applicable) [a]
Author(s)	
Year of publication	
Year of study conduct	
Country or countries of origin of data	
Sample size	
Type of sample	Public, other [describe]
Sample recruitment process	
Criteria for excluding respondents and/or observations reported?	Yes [describe], no
Mode of administration	Internet survey, computer-assisted personal interview, non-computer-assisted personal interview, focus group [b], self-completion paper survey [c], multiple modes [describe]
Summary of primary study objective(s)	e.g. 'To test for support for end of life prioritisation'
Was end of life (or a related term) mentioned explicitly in the study objectives?	Yes, no
Pilot reported?	Yes, no
Preference elicitation technique	Discrete choice experiment, other choice exercise, budget allocation, willingness-to-pay, ranking exercise, person trade-off, Q methodology, other
Perspective	Social decision-maker, own health, both
End of life definition	
Life expectancy without treatment attribute levels	
Life expectancy gain from treatment attribute levels	
Was disease labelled or named?	Yes [describe], no
Did the study examine whether quality of life-improving or life-extending treatments are preferred for end of life patients?	Yes, no / unclear
What were respondents choosing between (or choosing to do)?	e.g. 'Which of two patients to treat'
Was it possible to express indifference?	Yes, no, not reported
Were visual aids used?	Yes, no
Strength of preference examined at the individual respondent level?	Yes [describe], no / not reported
Number of tasks completed by each respondent	
Time taken to complete survey reported?	Yes [describe], no
Summary of finding: end of life vs. non-end of life	Evidence consistent with an end of life premium, evidence not consistent with an end of life premium, mixed or inconclusive evidence
Summary of finding: quality of life improvement vs. life extension	Quality of life improvement preferred, life extension preferred, not examined / reported
Other results of potential interest	
Other factors examined	
Impact of background characteristics reported? [d]	Yes [describe], no
Were qualitative data or explanatory factors sought?	Yes [describe], no
Was any reference made to age-related preferences? [e]	Yes [describe], no
Was any reference made to time-related preferences? [f]	Yes [describe], no

[a] Options with no observations are not indicated – for example, there is no telephone interviews option within the mode of administration field since no studies reported using that mode
[b] Defined as a group discussion guided by a moderator (or moderators) with individual completion of questionnaires (there were no observations of group discussions with group completion of questionnaires)
[c] Includes postal surveys and self-completion surveys administered within a group setting (but with no group discussion)
[d] For example, the authors may have reported that some respondent subgroups (e.g. females) were more likely than others to express support for an end of life premium
[e] For example, the authors may have reported evidence of support for an end of life premium for younger but not older patients
[f] For example, the authors may have attempted to control for the fact that health gains experienced now are valued more highly by individuals with a positive rate of time preference than health gains experienced later

2.3 Results

2.3.1 Searching for search terms

Table 2-2 lists the unique terms appearing in the abstracts of the three key papers, alongside a count of how many times each term appeared in the relevant abstract. Numbers written as digits (e.g. '10') have been excluded from the list.

Table 2-2. Terms appearing in the abstracts of the three key papers

Abel Olsen (2013)		Linley and Hughes (2013)		Shah *et al.* (2014)	
Term	Count	Term	Count	Term	Count
and	7	the	13	to	13
treatment	5	for	9	the	10
lifetime	4	and	7	of	8
a	3	to	5	be	6
age	3	criteria	4	that	6
in	3	health	4	for	5
remaining	3	of	4	in	5
that	3	societal	4	there	4
the	3	by	3	a	3
their	3	preferences	3	public	3
to	3	allocation	2	respondents	3
years	3	are	2	support	3
an	2	benefits	2	treatments	3
argument	2	cdf	2	and	2
expectancy	2	diseases	2	are	2
for	2	in	2	but	2
gain	2	medicines	2	choose	2
health	2	national	2	end-of-life	2
life	2	nice	2	evidence	2
of	2	nor	2	expressed	2
patients	2	not	2	giving	2
prognosis	2	or	2	health	2
support	2	prioritisation	2	if	2
without	2	proposed	2	is	2
analytical	1	resource	2	life	2
article	1	respondents	2	life-extending	2
as	1	system	2	may	2
asked	1	under	2	nice	2
be	1	vbp	2	not	2
characteristics	1	a	1	patients	2
choices	1	accepting	1	policy	2
depends	1	actual	1	preference	2
did	1	address	1	priority	2

Abel Olsen (2013)		Linley and Hughes (2013)		Shah *et al.* (2014)	
Term	Count	Term	Count	Term	Count
differed	1	adults	1	such	2
differences	1	allocate	1	them	2
disease	1	as	1	they	2
end-of-life	1	asking	1	this	2
entitled	1	assumed	1	treat	2
evidence	1	basis	1	we	2
fair	1	between	1	were	2
framework	1	but	1	which	2
from	1	cancer	1	would	2
how	1	children	1	preferences	2
hypothetical	1	choice-based	1	consensus	1
illustrate	1	clinical	1	account	1
implying	1	conducted	1	advice	1
increase	1	consequences	1	age	1
increasing	1	cost-effectiveness	1	answered	1
incremental	1	decisions	1	appraisal	1
incurable	1	determined	1	appraising	1
inequalities	1	did	1	appropriate	1
innings'	1	different	1	as	1
into	1	disadvantaged	1	asking	1
is	1	disease	1	assuming	1
little	1	drugs	1	between	1
loss	1	economic	1	both	1
make	1	effect	1	cannot	1
makes	1	empirical	1	certain	1
matter	1	end-of-life	1	clinical	1
month	1	england	1	committees	1
months	1	evidence	1	concerns	1
norwegian	1	excellence	1	conclude	1
not	1	experiment	1	considered	1
old	1	explore	1	consultation	1
on	1	fixed	1	control	1
our	1	fund	1	cost-effective	1
pairwise	1	funding	1	criteria	1
past	1	funds	1	current	1
patient	1	future	1	described	1
patients'	1	have	1	designed	1
population	1	higher	1	england	1
preferences	1	inappropriate	1	enough	1
preferential	1	including	1	even	1
presents	1	incremental	1	examines	1
prioritize	1	innovative	1	excellence	1
reduced	1	institute	1	expectancy	1
reveal	1	introduced	1	extension	1
sample	1	introduction	1	face-to-face	1
short	1	is	1	few	1
should	1	lacking	1	fifty	1
social	1	lead	1	find	1
splits	1	may	1	funds	1
strong	1	needs	1	general	1
such	1	new	1	has	1
terms	1	nine	1	higher	1
there	1	offered	1	however	1
this	1	on	1	hypothetical	1
total	1	other	1	improvement	1
untreated	1	others	1	indicated	1
value	1	over	1	indifference	1
was	1	patient	1	institute	1
who	1	perceived	1	insufficient	1
with	1	policies	1	interviewed	1

Abel Olsen (2013)		Linley and Hughes (2013)		Shah *et al.* (2014)	
Term	Count	Term	Count	Term	Count
year	1	population	1	interviews	1
		populations	1	into	1
		potential	1	issued	1
		premium	1	it	1
		pricing	1	its	1
		provided	1	july	1
		rare	1	little	1
		ratios	1	members	1
		recent	1	met	1
		reflect	1	minority	1
		reflecting	1	national	1
		rewarding	1	nice's	1
		robust	1	nontrivial	1
		service	1	normally	1
		severe	1	note	1
		significant	1	observed	1
		some	1	one	1
		special	1	opposite	1
		specific	1	or	1
		specified	1	other	1
		status	1	over	1
		substantial	1	patient	1
		support	1	prefer	1
		supported	1	quality-of-life	1
		surveys	1	questions	1
		these	1	recommend	1
		they	1	remaining	1
		this	1	results	1
		those	1	revealed	1
		treatments	1	scenarios	1
		types	1	scientific	1
		uk	1	service	1
		unmet	1	set	1
		used	1	shorter	1
		value-based	1	single	1
		values	1	six	1
		via	1	so	1
		we	1	some	1
		web-based	1	study	1
		were	1	substantial	1
		wider	1	suggest	1
		with	1	supplementary	1
				taken	1
				than	1
				time-related	1
				treatment	1
				two	1
				types	1
				unwillingness	1
				use	1
				various	1
				very	1
				was	1
				ways	1
				when	1
				whether	1
				whilst	1
				with	1

Table 2-3 shows the terms that appeared in at least two of the three key paper abstracts. Many of the commonly appearing words – such as articles (e.g. the), prepositions (e.g. of) and pronouns (e.g. this) – are of limited use in terms of informing literature search strategies. The asterisked words were deemed to be potentially relevant to the research question and the impact of adding these to the search was tested (see 2.2.1.1).

Table 2-3. Terms appearing in at least two of the three key paper abstracts

Term	Number of key paper abstracts this term appears in
with	3
to	3
this	3
the	3
support	3
preferences*	3
patient*	3
of	3
not	3
is	3
in	3
health*	3
for	3
evidence*	3
end-of-life*	3
as	3
and	3
a	3
were	2
we	2
was	2
types	2
treatments*	2
treatment*	2
they	2
there	2
that	2
such	2
substantial	2
some	2
service	2
respondents*	2
remaining	2
population*	2
patients*	2
over	2
other	2
or	2
on	2
nice	2
national	2
may	2
little	2
life	2
into	2
institute	2
incremental	2
hypothetical	2
higher	2

Term	Number of key paper abstracts this term appears in
funds	2
expectancy*	2
excellence	2
england	2
disease	2
did	2
criteria	2
clinical	2
but	2
between	2
be	2
asking	2
are	2

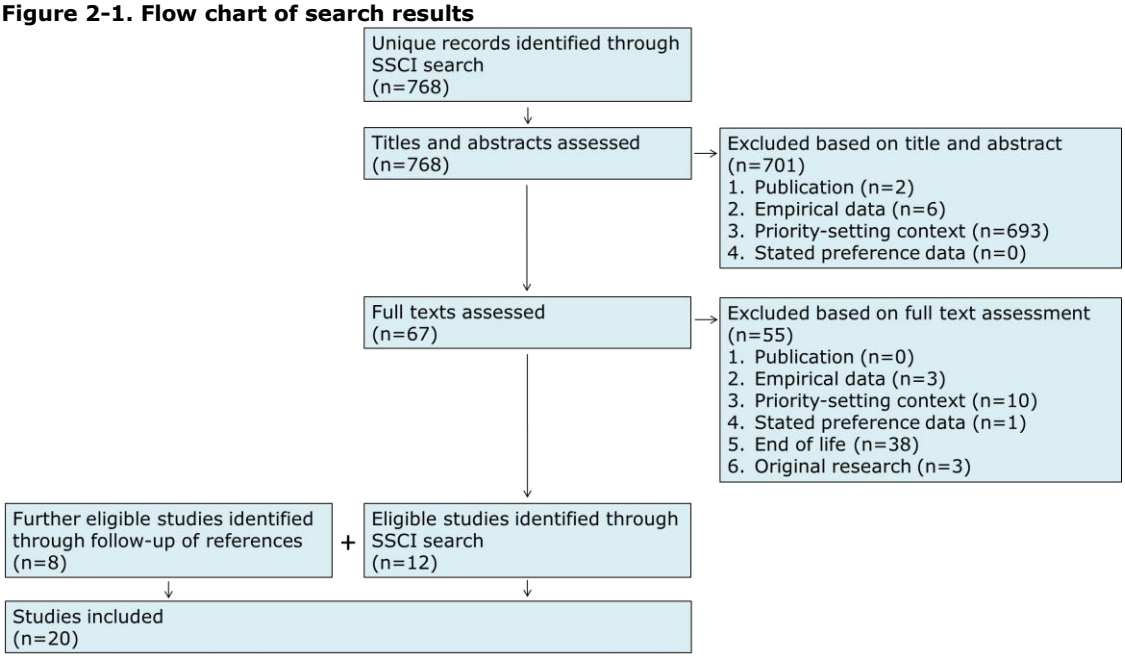
* Terms that were deemed by the author to be potentially relevant to the research question

2.3.2 Literature search output

The final (May 2016) SSCI search yielded 768 unique results (Figure 2-1). By comparison, the May 2014 search yielded 598 unique results. Following the review of titles and abstracts (in which inclusion criteria 1 to 4 were applied sequentially), 701 of these were excluded, mostly for failing to meet criterion 3. Commonly excluded articles at this stage included: studies about advance directives (living wills); studies of people's preferences for their own death and/or palliative care; studies focusing on the individual-level, bedside decision-making context; and health state valuation studies. Both of the articles excluded for failing to meet criterion 1 were published in German.

Following a review of the full texts of the remaining 67 records (in which inclusion criteria 1 to 6 were applied sequentially), a further 55 were excluded. Commonly excluded articles at this stage reported public preferences regarding the prioritisation of health care resources based on severity (amongst other criteria) but did not define severity in terms of life expectancy, or did not report the results in such a way that preferences regarding life expectancy could be inferred.

Figure 2-1. Flow chart of search results



The reference lists of the articles whose full texts were reviewed identified a further eight articles that were relevant to the research question but had not been picked up by the SSCI search. For example, two of these articles did not include the term *health* in their titles, abstracts or keywords. The additional articles met all six of the criteria for inclusion. The initial review had included four articles that were known to the author and were considered relevant to the research question, but had been published after the SSCI search was conducted. These four articles were all identified in the updated search.

In cases where an article described a large study comprising multiple sub-studies with distinct methods and/or samples (e.g. Baker *et al.*, 2010a), only the data for the sub-studies that were relevant to the research question were extracted.

2.3.3 Description of included studies

The included articles (Table 2-4) were published between 2000 and 2015, with the majority conducted and published after NICE issued its supplementary advice on end of life in January 2009. Ten of the studies (50%) used a solely UK-based sample, with the other studies originating elsewhere in Europe and in Australia, Canada, Japan, South Korea and the United States. Two studies included multi-country samples (Pennington *et al.*, 2015; Shiroiwa *et al.*, 2010). The distribution of key variables across the 20 articles is shown in

Table 2-5. Full details are available in Appendix 3.

Table 2-4. Summary of included studies (n=20)

Record	Authors (date)	Country	Sample size (type)	Method [a]	Mode of administration	Summary of primary study objective(s)
1	Abel Olsen (2013)	NOR	503 (public)	Choice	Internet survey	To test for support for end of life prioritisation and the fair innings approach
2	Baker *et al.* (2010a)	UK	587 (public)	DCE	Computer-assisted personal interview	To test for support for multiple prioritisation criteria
3	Dolan and Cookson (2000)	UK	60 (public)	Choice	Focus groups (individual responses)	Qualitative examination of support for multiple prioritisation criteria
4	Dolan and Shaw (2004)	UK	23 (public)	Choice	Focus group (individual responses)	To test for support for multiple prioritisation criteria
5	Dolan and Tsuchiya (2005)	UK	100 (public)	Choice; ranking	Individual self-completion survey (completed in group setting)	To compare support for prioritisation according to age vs. prioritisation according to severity/life expectancy
6	Lim *et al.* (2012)	ROK	800 (public)	DCE	Internet survey	To test for support for multiple prioritisation criteria
7	Linley and Hughes (2013)	UK	4,118 (public)	Budget allocation	Internet survey	To test for support for multiple prioritisation criteria
8	McHugh *et al.* (2015)	UK	61 ('data-rich' individuals) [b]	Q method	Non-computer assisted personal interview	Qualitative examination of societal perspectives in relation to end of life prioritisation
9	Pennington *et al.* (2015)	Multiple	17,657 (public)	WTP	Internet survey	To compare WTP for different types of QALY gain
10	Pinto-Prades *et al.* (2014)	SPA	813 (public)	WTP; PTO	Computer-assisted personal interview	To test for support for end of life prioritisation and to compare support for life extensions vs. quality of life improvements
11	Richardson *et al.* (2012)	AUS	544 (public)	Other	Multiple modes: Internet survey and self-completion survey (postal)	To test a technique for measuring support for health-maximisation and health sharing
12	Rowen *et al.* (2016a)	UK	3,669 (public)	DCE	Internet survey	To test for support for multiple prioritisation criteria
13	Rowen *et al.* (2016b)	UK	371 (public)	Choice	Multiple modes: Internet survey and	To test for framing and mode of administration effects in the elicitation of preferences regarding burden of illness

Record	Authors (date)	Country	Sample size (type)	Method [a]	Mode of administration	Summary of primary study objective(s)
					non-computer assisted personal interview	
14	Shah *et al.* (2014)	UK	50 (public)	Choice	Non-computer assisted personal interview	To test for support for end of life prioritisation
15	Shah *et al.* (2015a)	UK	3,969 (public)	DCE	Internet survey	To test for support for end of life prioritisation
16	Shiroiwa *et al.* (2010)	Multiple	5,620 (public)	WTP	Internet survey	To obtain the monetary value of a QALY (in six countries)
17	Shiroiwa *et al.* (2013)	JPN	2,283 (public)	WTP	Internet survey	To obtain the monetary value of a QALY
18	Skedgel *et al.* (2015)	CAN	595 (public); 61 (decision-makers)	DCE	Internet survey	To test for support for multiple prioritisation criteria
19	Stahl *et al.* (2008)	USA	623 (public)	Choice	Internet survey	To test for support for multiple prioritisation criteria
20	Stolk *et al.* (2005)	NLD	65 (students, researchers, health policy makers)	Choice	Non-computer assisted personal interview	To test for support for multiple approaches to priority-setting

[a] Choice = choice exercise that did not include design or analysis methods associated with the DCE technique; DCE = discrete choice experiment; PTO = person trade-off; WTP = willingness-to-pay

[b] Made up of 59 data-rich individuals with different types of experiences or expertise in end of life in a professional and/or personal capacity (e.g. researchers, pharmaceutical industry employees, patient group representatives, religious group representatives, clinicians, people with experience of terminal illness in family members), plus two 'meta-respondents' representing the views of 250 general public respondents.

Table 2-5. Distribution of key variables (n=20)

Variable	Frequency	%
Year of study publication		
- Prior to 2009	5	25%
- 2009 onwards	15	75%
Year of study conduct [a]		
- Prior to 2009	7	35%
- 2009 onwards	13	65%
Sample size		
- 1-99	5	25%
- 100-999	9	45%
- 1,000+	6	30%
Perspective		
- Own health	2	10%
- Social decision-maker	16	80%
- Both	2	10%
Method / preference elicitation technique		
- Discrete choice experiment	5	25%
- Other choice exercise	7	35%
- Budget allocation	1	5%
- Q methodology	1	5%
- Willingness-to-pay	3	15%
- Person trade-off and willingness-to-pay [b]	1	5%
- Ranking exercise and other choice exercise [c]	1	5%
- Other	1	5%
Mode of administration		
- Internet survey	10	50%
- Computer-assisted personal interview	2	10%
- Non-computer-assisted personal interview	3	15%
- Focus group	2	10%
- Self-completion paper survey (completed in group setting)	1	5%
- Multiple modes [d]	2	10%
Disease labelled?		
- No	16	80%
- Yes – choice between several named diseases	1	5%
- Yes – choice between treatments for a single named disease	3	15%
Shortest life expectancy presented		
- 0mths (i.e. imminent death)	4	20%
- 0mths < LE ≤3mths	6	30%
- 3mths < LE ≤ 12mths	6	30%
- 12mths < LE	3	15%
- No length specified	1	5%
Possible to express indifference?		
- Yes	10	50%
- No	9	45%
- Not reported / unclear	1	5%
Visual aids used?		
- Yes [e]	10	50%
- No	10	50%
Strength of preference examined at the individual respondent level?		
- Yes	9	45%
- No	10	50%
- Not reported / unclear	1	5%
Qualitative data or explanations for choices sought?		
- Yes	8	40%
- No / not reported	12	60%
Impact of background characteristics		
- At least one characteristic found to be associated with preferences	5	25%
- No characteristics found to be associated with preferences	6	30%
- Not reported	9	45%
Any reference to age-related preferences?		
- Yes	12	60%
- No	8	40%
Any reference to time-related preferences?		
- Yes – an attempt was made to control for or analyse time-related preferences	3	15%
- Yes – time-related preferences were mentioned but not controlled for	3	15%
- No	14	70%

Variable	Frequency	%
Overall finding: end of life vs. non-end of life		
- Consistent with an end of life premium	7	35%
- Not consistent with an end of life premium	9	45%
- Mixed or inconclusive evidence	4	20%
Overall finding: quality of life-improving vs. life-extending end of life treatments	2	10%
- Quality of life improvement preferred	1	5%
- Life extension preferred	1	5%
- Mixed or inconclusive evidence	16	80%
- Not examined / reported		

[a] Not always reported – in some cases this was inferred based on the year of study publication; in other cases clarification was sought by means of personal communication with authors
[b] Separate methods – all respondents completed tasks using both methods
[c] Hybrid method – all respondents were asked first to choose which of six patient groups to treat, and then to rank the six patient groups in order of preference
[d] Internet survey and self-completion paper survey; internet survey and non-computer-assisted personal interview
[e] One study is counted as a study that used visual aids on the basis that visual aids were used in the majority of study arms (and for the majority of respondents)

2.3.4 Methods used to elicit preferences

Thirteen of the 20 studies (65%) elicited preferences using some form of choice exercise whereby respondents were presented with multiple hypothetical patients (or patient groups) and were asked which they thought should be treated. In most cases the tasks involved pairwise choices, though Dolan and Shaw (2004) and Dolan and Tsuchiya (2005) both asked respondents to choose between six alternatives. Five of the choice exercise studies explicitly applied the DCE method as defined by Carson and Louviere (2011) – that is, an approach in which choices are made between discrete alternatives where at least one attribute is systematically varied in such a way that information related to preference parameters of an indirect utility function can be inferred.

A related approach, budget allocation, allows respondents to indicate the strength of their preference by specifying how funding should distributed among the candidate beneficiaries. The results of this method can be simplified by reporting, for example, whether respondents gave the majority of the budget to one group or another, or opted for an equal allocation between the groups. Indeed, the sole budget allocation study (Linley and Hughes, 2013) reported their results in this way. Respondents had 11 different distributions of funding to choose from but the authors collapsed the results into three categories (as above), making them comparable with studies which used pairwise choice tasks with an indifference option (e.g. Shah *et al.*, 2014).

Most of the studies used methods that are well-established in the field of health care preference elicitation (Ryan *et al.*, 2001). A more novel approach, which combined elements of the budget allocation and choice exercise techniques, was used in one study (Richardson *et al.*, 2012). Respondents were asked to allocate a

set budget to one of the four patients (all of whom were the same age and faced immediate death without treatment), which would have the effect of extending their lives by 12, 8, 6 or 4 years, respectively. After allocating the first budget, they were then given a second budget (of the same size and with the same life-extending effects) to allocate in addition to the first. The procedure was repeated 30 times. Respondents' allocations gave an indication of whether they sought to maximise the number of years gained or to sacrifice overall gains by giving priority to the patient with the shortest life expectancy.

Another less established approach (in the field of health economics, at least) – Q methodology (Watts and Stenner, 2012) – was used in one study (McHugh *et al.*, 2015). Q methodology combines qualitative and quantitative methods to study people's subjective opinions, values and beliefs (Baker *et al.*, 2006). Respondents were presented with 49 statements describing views relating to the provision of end of life treatments. Following a structured process, they were asked to sort and position the statements on a response grid depending on whether they agreed with, disagreed with or were neutral towards them. They were then asked to articulate their views and to comment on statements that had been placed in the extremes of the grid. The researchers used factor analysis to identify underlying patterns in the resulting 'Q sorts'.

Four studies employed the willingness-to-pay method (Shiroiwa *et al.*, 2010; Shiroiwa *et al.*, 2013; Pinto-Prades *et al.*, 2014; Pennington *et al.*, 2015), in which respondents were asked whether and how much they would be willing to pay, from their own pocket, for a given improvement in health or life extension – or in the case of Pinto-Prades *et al.* (2014), for a specified chance of improvement. Respondents were generally expected to take an 'own health' perspective (i.e. to imagine that they were the beneficiaries of the treatment on offer) when completing the willingness-to-pay tasks. The other studies employed a 'social decision-maker' perspective whereby respondents were expected to make choices that they considered most appropriate and acceptable for society rather than those guided purely by self-interest. One study employed both an own health perspective, in willingness-to-pay tasks, and a social decision-maker perspective, in person trade-off tasks (Pinto-Prades *et al.*, 2014). Another study examined respondents' willingness-to-pay for life extensions not only for themselves but also for a family member (via an out-of-pocket payment) and for an unidentified member of society (via a tax increase) (Shiroiwa *et al.*, 2010).

One study compared two operationalisations of the social decision-maker perspective, asking half of the respondents to adopt the role of a decision-maker

and assigning the other half to a 'veil of ignorance' condition (Dolan and Cookson, 2000). In the Q methodology study, the vast majority of statements presented were framed in a manner consistent with a social decision-maker perspective, though a few referred to the respondent's own health or situation – for example, "I wouldn't want my life to be extending just for the sake of it – just keeping breathing is not life" (McHugh *et al.*, 2015).

Thirteen studies (65%) used modes of administration that required respondents to complete the tasks without an interviewer or moderator present to provide guidance. With one exception (Baker *et al.*, 2010a), the DCE studies were all administered via internet surveys, most likely due to the ease of obtaining large samples with this mode. There has been a shift towards computer-based survey administration over time – the review included only two studies published since 2005 which did not use either an internet survey or computer-assisted personal interview approach.

Visual aids were used by 10 studies (50%), including all of the DCE studies. The same number of studies permitted respondents to express indifference between or assign equal value to the alternatives presented. Eleven studies (55%) reported that their design had been informed by piloting.

In each study, with the exception of the Q methodology study, the size of the health gain was controlled for either in the design (e.g. by presenting equal-sized gains for all candidate recipients) or in the analysis.

2.3.5 Samples

Most of the studies used general public samples, though the extent to which the samples were representative of the relevant populations was mixed. McHugh *et al.* (2015) used a purposive sample comprising data-rich individuals (that is, individuals expected to have 'rich, strong and different views' on the topic) with different types of experiences or expertise in end of life in a professional or personal capacity. Skedgel *et al.* (2015) surveyed a small number of decision-makers (n=61; out of a total sample of 656 respondents) with the aim of contrasting their responses with those of the general public. Stolk *et al.* (2005) used a convenience sample consisting of students, researchers and health policy makers – all of whom had some level of expertise in the topic of health care priority-setting.

The samples ranged from 23 individuals recruited from a single small city (Dolan and Shaw, 2004) to 17,657 individuals recruited from nine different countries (Pennington *et al.*, 2015). The six largest-sample studies (n≥1,000) were all

administered over the internet and recruited respondents from panels managed by market research agencies.

2.3.6 End of life definitions

Eight articles (40%) explicitly mentioned end of life, or some synonym for end of life, in the stated study objectives. Of the remaining studies, some included end of life amongst several prioritisation criteria examined (e.g. Linley and Hughes, 2013), whilst others sought to answer an altogether different research question but happened to provide evidence relevant to end of life-related preferences indirectly (e.g. Richardson *et al.*, 2012). In the latter cases, preferences regarding end of life were inferred by extracting the results that could be used to draw conclusions about the values of a given gain for patients with different life expectancies (occasionally making calculations beyond those presented in the journal articles as necessary). End of life was most commonly presented in terms of patients' 'life expectancy' or 'remaining life years' if they did not receive the treatment, health care or transplant on offer. Other terms used included 'future years', 'urgency', 'fatal disease' and 'imminent death'.

A wide range of levels for the 'life expectancy without treatment' attribute (where applicable) was used. Some studies, none of which explicitly set out to examine preferences related to end of life, asked respondents to consider scenarios where patients would die immediately in absence of treatment, which meant in effect that their life expectancy without treatment was zero. In two studies (Stolk *et al.*, 2005; Baker *et al.*, 2010a), information on the patients' life expectancy was not presented directly but could be calculated using the attributes that *were* included.

All but three of the studies presented at least one alternative in which the patient or patient group would live for less than two years without treatment, which would make them potentially eligible for special consideration under NICE's criteria (NICE, 2009c). The first exception is Dolan and Cookson (2000), whose only question relevant to this review involved choosing between two patient groups who would live for 10 or 30 years without treatment (they included other questions in which the life expectancies were shorter but did not differ between the patient groups, hence no end of life versus non-end of life comparison could be made). The second exception is Rowen *et al.* (2016b), whose only question relevant to this review involved choosing between two patient groups who would live for five or 10 years without treatment. The third exception is McHugh *et al.* (2015), whose method did not involve choices between patients with specified life expectancies, and whose statements did not mention specific life expectancy values.

2.3.7 Comparators and other attributes examined

In the majority of studies, the key comparison – at least for the purposes of this review – was between an alternative describing a short, fixed amount of remaining life without treatment and one or more alternatives describing longer, fixed amounts of remaining life without treatment. Three studies, all of which applied the willingness-to-pay method, used different types of comparators (Shiroiwa *et al.*, 2013; Pinto-Prades *et al.*, 2014; Pennington *et al.*, 2015). These studies all included scenarios involving temporary quality of life losses, and sought respondents' willingness-to-pay to avoid those losses. One of the three studies also included a scenario involving a life extension at the end of the respondent's own stated life expectancy, and another involving spending time in a coma (Pennington *et al.*, 2015).

While several studies included attributes relating to quality of life gains, only three explicitly tested and reported whether respondents preferred quality of life improvements or life extensions for end of life patients (Pinto-Prades *et al.*, 2014; Shah *et al.*, 2014; Shah *et al.*, 2015a). Other studies collected the data required to make such comparisons possible but did not focus on quality of life in the published articles.

Nine studies (45%) purposely included information about age, thereby providing evidence on interactions between respondents' preferences regarding age and regarding end of life. One study (5%) attempted to control for time-related preferences by including questions designed to identify whether any observed preference for treating patients with shorter life expectancy is driven by a preference for the benefits of treatment to occur sooner rather than later (Shah *et al.*, 2014).

2.3.8 Findings of the studies

2.3.8.1 Evidence consistent with an end of life premium

Seven studies (35%) report evidence of support for placing greater weight on a unit of health gain for patients with relatively short life expectancy than on that for other types of patients. Their findings are summarised briefly below (presented in chronological order).

Stahl *et al.* (2008) report that respondents preferred treating the patient who was closer to death until the difference in life expectancy was less than 1.1 months (beyond which they showed no preference for the patient with shorter life expectancy). They also report that when one patient was set to gain a shorter life

extension than another, the former needed to have a shorter life expectancy without treatment in order to be given priority overall (up to a threshold).

Shiroiwa *et al.* (2010) report that in all six countries examined, higher willingness-to-pay values were observed in scenarios where respondents had zero years of life expectancy than in scenarios where they had five years of life expectancy.

Lim *et al.* (2012) report that higher priorities were given to patients with less remaining life, noting that that respondents overall were willing to give up a 0.39 QALY gain in order to treat the patient whose life expectancy without treatment was one level (usually five years) lower.

Pinto-Prades *et al.* (2014) report that six- or 18-month life extensions for end of life patients were valued more highly than temporary quality of life improvements for non-end of life patients that were equivalent in terms of the number of QALYs gained. They note that this result was observed in both the willingness-to-pay and the person trade-off surveys, though the patterns of responses differed across the two methods.

Rowen *et al.* (2016a) report results that showed support for an end of life premium across different regression models, with evidence of a preference for treating patients with shorter life expectancy without treatment. However, the responses to their follow-up (attitudinal, non-choice-based) questions appear to contradict this finding.

Shah *et al.* (2014) report that the majority of respondents chose to give a six-month life extension to the patient with one year left to live without treatment rather than to the patient with 10 years left to live without treatment. However, they also noted that a non-trivial minority of respondents expressed the opposite preference.

Pennington *et al.* (2015) report that the mean and median willingness-to-pay values for one QALY worth of life extension achieved in the scenario of 'imminent, premature death from a life threatening disease' were considerably larger than those for an equal-sized gain achieved at the end of respondents' self-predicted life expectancy.

2.3.8.2 Evidence not consistent with an end of life premium

Nine studies (45%) report evidence that people do *not* wish to place greater weight on a unit of health gain for patients with relatively short life expectancy than on that for other types of patients.

Dolan and Shaw (2004) report that the majority of respondents chose to give priority to the patient with the longest life expectancy without a kidney transplant and who stood to gain the most from receiving the transplant. When it was later revealed that the end of life patient was the oldest of the six candidate recipients, none of the respondents chose to give the transplant to that patient.

Dolan and Tsuchiya (2005) report that respondents priority ranked end of life patients lower than corresponding non-end of life patients for all levels of age and past health. They also note that the coefficient for future years (life expectancy without treatment) as a main effects variable was not statistically significant.

Stolk *et al.* (2005) report priority rankings (based on respondents' choices in paired comparison tasks) of 10 conditions that correlated poorly and non-significantly with the theoretical ranking implied by a 'priority to shorter life expectancy' approach. Other theoretical rankings (severity, fair innings, proportional shortfall) were all significantly correlated with the observed ranking. Respondents were less concerned about life-threatening conditions for the elderly than prospective health theories that ignore the past (i.e. age) would have predicted.

Abel Olsen (2013) reports evidence of strong support for the fair innings argument, noting that respondents' choices were not affected by differences in patients' remaining lifetime without treatment.

Linley and Hughes (2013) report that, when faced with a choice between treating one patient group with a life expectancy of 18 months and another patient group with a life expectancy of 60 months, about two-thirds of respondents opted not to allocate more resources to the end of life group. The most popular choice was to allocate an equal amount of funding to both groups.

Shiroiwa *et al.* (2013) report that the proportions of respondents willing to pay an initial bid value for gains worth 0.2 or 0.4 QALYs were consistently lower in end of life scenarios than in non-end of life scenarios. Further, the average willingness-to-pay per QALY values observed in the end of life scenarios were generally lower than in the non-end of life scenarios.

Shah *et al.* (2015a) report a statistically significantly negative coefficient for the life expectancy without treatment variable, but noted that it was very small in magnitude compared to the health gain coefficients and had very little impact on the choices made by respondents. An end of life dummy variable defined purely in terms of life expectancy without treatment was found to have a small and non-significant coefficient.

Skedgel *et al.* (2015) report evidence of statistically significant and negative (positive) welfare effects associated with prioritising patients with the shortest (longest) level of initial life expectancy.

Rowen *et al.* (2016b) report an approximately equal split between choosing to treat a patient group with a life expectancy of five years and choosing to treat another patient group with a life expectancy of 10 years. Tests of association conducted by the authors indicate that this result did not depend on the mode of administration, wording of the question, or use of visual aids.

2.3.8.3 Studies reporting mixed or inconclusive evidence

Four studies (20%) reported evidence that cannot easily be interpreted as being clearly consistent or inconsistent with an end of life premium. This was either because of heterogeneous preferences or because the observed results were not sufficiently robust.

Dolan and Cookson (2000) report that when asked to choose between giving a 10-year life extension to one patient group with 10 years of life expectancy without treatment and another with 30 years of life expectancy, 2% of respondents chose the latter; 50% chose the former; and 48% gave the same priority to both groups.

Baker *et al.* (2010a) assessed preferences for different scenarios relative to a reference scenario of treating 40 year old patients expected to die at 60 years with a 0.7 quality of life loss without treatment. They report that in scenarios which were purely life-saving (i.e. involving immediate death without treatment), a preference was observed for treating patients aged 10 years relative to the reference scenario (controlling for the size of QALY gain). For other ages (1, 40 or 70 years), the reference scenario was preferred to the life-saving treatments. Similarly, life-saving treatments for 10 year old patients were preferred to treatments (offering the same QALY gains) for 10 year old patients who would not die immediately if left untreated, whilst the opposite was observed for patients of other ages.

Richardson *et al.* (2012) report that the majority of respondents did not behave in a QALY-maximising manner, with 69% allocating one of their first four budgets to the patient who stood to gain least (a four-year life extension) rather than giving that budget to the patient who stood to gain most (12-year life extension). The authors note that the average respondent allocated resources in such a way that 62.6% of possible gains in life years were achieved, with 37.4% of gains sacrificed to achieve sharing. In their regression models, life expectancy is a dominating variable – across all choices, the greater a given patient's life expectancy, the smaller the probability of that patient receiving resources (i.e. further life extensions).

McHugh *et al.* (2015) identified three 'factors' (shared perspectives) in their analysis. The first factor describes the view that society's interests are best served by seeking to maximise population health, and that "terminal illness should not be treated as a special case" (p.9). The second factor emphasises patient choice and the right to life-extending treatment for patients who want it, though this right may apply to non-end of life as well as end of life conditions. The third factor permits cases where special value is placed on extending the life of end of life patients, but this value is not unconditional and must be weighed up against opportunity costs. The findings demonstrate the 'plurality of views' within society and the authors highlight the problems associated with determining policy based on simple majority votes.

Table 2-6 compares the distribution of selected variables of interest among studies that report evidence consistent with an end of life premium with those among studies that do not.

Table 2-6. Distribution of selected variables, by overall study finding

Variable	Evidence consistent with an end of life premium	Evidence not consistent with an end of life premium
Country		
- UK	2	5
- Europe (non-UK)	2	2
- Rest of the world [a]	3	2
Method [b]		
- DCE	2	2
- Other choice exercise	2	4
- Willingness-to-pay	3	1
- Other	1	2
Mode of administration [c]		
- Internet survey	5	6
- Other	2	4
Possible to express indifference?		
- Yes	5	3
- No or not reported	2	6
Visual aids used? [d]		
- Yes	5	3
- No or not reported	2	7

[a] Includes a multi-country study conducted in Australia, Japan, South Korea, Taiwan, UK and USA. Counted as a 'Rest of the world' study because the UK sample comprised less than 20% of the total sample.
[b] Study combining person trade-off and willingness-to-pay methods counted as two studies since separate results are reported for both. Study combining ranking exercise and other choice exercise counted as one study since this is considered to be a single hybrid method.
[c] Study combining internet survey and non-computer-assisted personal interview modes of administration counted as two studies since separate results are reported for both.
[d] Study combining visual aid and no visual aid arms counted as two studies since separate results are reported for both.

2.3.8.4 Other findings of relevance

Most of the studies did not examine or report explicitly whether quality of life improvements or life extensions for end of life patients were preferred, though in some cases it would have been possible to examine this given the nature of the data collected. Two studies reported that respondents favoured quality of life improvements (Pinto-Prades *et al.*, 2014; Shah *et al.*, 2014); and one study reported that respondents favoured life extensions (Shah *et al.*, 2015a) – controlling for the size of health gain in all cases.

The majority of studies included patient age in the study design. In some cases age was one of several prioritisation criteria being examined; in other cases, the researchers were seeking to examine whether respondents' end of life-related preferences were influenced by the ages of the patients. The findings of two studies suggest that respondents become less concerned about the number of remaining life years when the patients in question are relatively old (Dolan and Shaw, 2004; Stahl *et al.*, 2008). One study did not find that concern about age is a motivating factor for giving priority to the treatment of end of life patients (Shah *et al.*, 2014), though the range of ages presented was narrow (nine years). Several studies reported evidence that respondents gave priority to younger patients, often without making an explicit link between age-related preferences and end of life-related preferences (Dolan and Cookson, 2000; Dolan and Tsuchiya, 2005; Stolk *et al.*, 2005; Baker *et al.*, 2010a; Abel Olsen, 2013; Skedgel *et al.* 2015).

Time-related preferences were mentioned in only a few of the studies. One study reported evidence that patients who have only just learned their prognosis are given priority over those who have known about their prognosis for some time, controlling for life expectancy (Shah *et al.*, 2014).[13] Another study interpreted differences between willingness-to-pay values in end of life and non-end of life scenarios in terms of time preference, and used the data to estimate discount rates (Shiroiwa *et al.*, 2010). Three studies acknowledged that their findings may have been influenced by respondents' time preference or that applying a positive discount rate in the analysis would have led to slightly (albeit not qualitatively) different results (Richardson *et al.*, 2012; Pennington *et al.*, 2015; Shah *et al.*, 2015a).

Two studies reported evidence that older respondents were more likely than average to make choices based on patients' life expectancy without treatment (Dolan and Tsuchiya, 2005; Stahl *et al.*, 2008). One of the willingness-to-pay

[13] Similar analyses were also conducted by Shah *et al*. (2015b) but were not reported in the journal article and were therefore not included in the review. Full details are available in Chapter 5 of this thesis.

studies reported that older age was associated with lower valuation for life extensions in the own terminal illness scenario (Pennington *et al.*, 2015). Other background characteristics found to be associated with respondents' priority-setting preferences were: education (Dolan and Tsuchiya (2005) and Shiroiwa *et al.* (2010) found that respondents with higher levels of education were more likely to place greater value on end of life treatments); employment status (Dolan and Tsuchiya (2005) found that respondents in employment were less likely to prioritise the treatment of end of life patients); health status (Pennington *et al.* (2015) found that respondents in better health gave higher valuations for life extensions at their own end of life); health history of family members (Stahl *et al.* (2008) found that respondents with transplant recipients in their family tended to prioritise transplants for patients with poor quality of life over those for patients with short life expectancy unless remaining life was extremely short, i.e. less than one month); and household income (Shiroiwa *et al.* (2010) found that respondents with higher household income levels gave higher willingness-to-pay values for life extensions at the end of life). However, the majority of studies either did not observe any associations between background characteristics and preferences or did not report any such analysis. McHugh *et al.* (2015) found that none of the academic researchers in their sample helped to define the shared account most closely related to support for an end of life premium, though the authors warn against making generalisations based on qualitative samples.

2.4 Discussion

Twenty empirical studies that inform the research 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 were identified and reviewed. The number of studies addressing this topic has been growing – several were initiated following (and refer explicitly to) the issuing of NICE's supplementary advice on end of life treatments in January 2009. Many of the studies originated in the UK, which is unsurprising given the policy interest in NICE (an agency which make recommendations on the use of health technologies in England) and the country's tradition of contributing to the literature on empirical ethics in health (Green, 2011). Most of the studies reviewed used a preference elicitation technique that can be described as a 'choice exercise', with an increasing number specifically applying the DCE method. This reflects the growing popularity of the method in applied health economics research (Ryan *et al.*, 2008; de Bekker-Grob *et al.*, 2012; Clark *et al.*, 2014), particularly in the field of health care priority-setting (Whitty *et al.*, 2014). DCEs are considered to enjoy a strong theoretical basis (Lancsar and

Donaldson, 2005) and there is evidence that the method is feasible, flexible and capable of presenting choices that are relevant to respondents (Louviere *et al.*, 2000; Ryan and Gerard, 2003).

The primary finding of the review is that the existing evidence is mixed, with seven studies that report evidence consistent with a premium for end of life treatments and nine studies that do not. To the best of the author's knowledge, no comparable up-to-date reviews are available. Two unpublished reviews of social value judgements in health care priority-setting, including those related to end of life, have been undertaken. One did not identify any relevant studies (Green, 2011); the other described the evidence base as 'not strong' (Keetharuth *et al.*, 2011). In comparison to those reviews, this review was more successful in identifying relevant studies, in part because many of those studies have been undertaken and published in the last few years. Further, more specific and comprehensive approaches to searching for articles and extracting the relevant data were used in this review.

Reviews of severity-related preferences more generally have been able to reach more decisive conclusions – Shah (2009) and Nord and Johansen (2014) both report an overall preference for giving higher priority to those who are severely ill – but as mentioned above the studies reviewed typically focused on severity in terms of quality of life, not length of life. Comparing the findings of the reviews of severity with those of the present review suggests that people are more likely to be concerned about treating patients with poor quality of life than with treating patients with short life expectancy. However, this supposition is not supported by individual studies that examined both simultaneously – Stahl *et al.* (2008), Shah *et al.* (2015a) and Rowen *et al.* (2016a) all report stronger support for giving priority to treating patients with relatively short life expectancy than to treating those with relatively poor quality of life, controlling for the size of health gain.

Whether quality of life improvements or life extensions were preferred for end of life patients was also examined. Although several studies collected data on both types of health gain, only three reported their results so as to make this comparison possible.[14] Again, the evidence is mixed, with two studies reporting evidence of an overall preference for quality of life improvement over life extension, and one study reporting the opposite. It is noteworthy that the current NICE policy involves giving greater weight to life-extending but not to quality of life-improving

[14] It should be noted that the primary focus of this review was whether gains for end of life patients are given more weight than gains for non-end of life patients, hence studies that focused only on end of life without a non-end of life comparator were excluded based on inclusion criterion 5 (see 2.2.2). Some of the excluded studies may have contained relevant information about whether quality of life improvements or life extensions are preferred in an end of life context.

treatments for those at the end of life. There is little evidence to suggest that such a policy is consistent with public preferences.

The overall findings of studies were summarised by assigning each to one of three categories: (1) consistent with an end of life premium; (2) not consistent with an end of life premium; and (3) mixed or inconclusive evidence. In absence of a clear definition of what counted as 'support', this exercise involved a degree of subjective judgement. It is rarely the case in stated preference studies that a unanimous preference is observed. There is usually a split in opinion, and a judgement then needs to be made about whether the minority view is held by a sufficiently large number of respondents (or held sufficiently strongly) so as to conclude that the evidence is inconclusive overall. As far as possible, the study authors' own conclusions were used as a guide. This was not always possible, since some studies did not set out to examine end of life-related preferences directly and further subjective interpretation of the reported results was required. In cases where there was uncertainty about the conclusions of a given study, the corresponding author was contacted to check that they agreed with the proposed summary and categorisation of their findings.

The heterogeneity of preferences held by the general population is highlighted by McHugh *et al.* (2015), who identified three distinct shared perspectives in their data. If most or all of their respondents had expressed agreement with statements indicating clear support for prioritising treatments for end of life patients, the authors' methods of analysis would have allowed a shared viewpoint consistent with an end of life premium to have emerged (McHugh, N., 2016, personal communication, 26 September). However, the data indicate that the views held by respondents were in fact more varied and nuanced than that. Other studies similarly identified multiple subgroups within their samples whose response patterns imply very different views about the value of end of life treatments (e.g. Pinto-Prades *et al.*, 2014; Shah *et al.*, 2015a). Given these findings, it is perhaps not surprising that this review has been unable to establish whether or not the overall evidence available in the literature is consistent with an end of life premium.

Majoritarian decision rules are common in politics and policy making, with most elections and referendums in modern western democracies being decided by majority rule. However, such approaches are criticised for failing to achieve outcomes that represent the views of all sections of society in a representative manner (Mill, 1861). A hypothetical example of a study that would be problematic to categorise based on majority rule is one in which a slight (but statistically significant) majority of respondents express *weak* support for an end of life

premium and a sizeable minority *strongly* disfavour an end of life premium. Many of the studies in this review did not examine strength of preference at the individual respondent level and were not designed in such a way that nuances and caveats regarding respondents' stated preferences could be captured. The normative basis for specifying a measure of average or overall preference in social choices is unclear – in the context of aggregating preferences regarding health states, Devlin *et al.* (2017) conclude that there are no strong grounds for favouring any one approach.

Table 2-6 shows how studies that report evidence consistent with an end of life premium compare to those that do not. The number of studies included in the review is insufficient to permit meaningful testing of statistical associations, so any trends observed should be interpreted with caution. Nevertheless, there is weak evidence that studies were more likely to report evidence consistent with an end of life premium if they: used the willingness-to-pay method; allowed indifference to be expressed; or used visual aids. Each of these variables is discussed in turn below.

2.4.1 Choice of method

Most of the studies in this review asked respondents to adopt a social decision-maker perspective – that is, they were asked to consider questions typically of concern to a health care decision-maker (such as whether one patient group or another should receive higher priority in the face of scarce shared resources) and to answer those questions based on what they consider to be appropriate and acceptable for society. The respondent (acting as decision-maker for the purpose of the study) would not necessarily expect to benefit personally from their choices. The four studies that used the willingness-to-pay approach, on the other hand, generally asked respondents to adopt an individual or own health perspective – that is, they were asked how much they would pay (from their own pocket) for a given improvement in their own health. This method is consistent with the welfarist view that confines the evaluative space to individual utility only – the 'goodness' of a policy can be judged solely on the basis of the utility gains and losses achieved by individuals affected by that policy (Brouwer *et al.*, 2008).

Three of the four willingness-to-pay studies report evidence consistent with an end of life premium, based on higher average willingness-to-pay values for a life extension in an end of life scenario than for a similar gain (e.g. worth the same number of QALYs) in a non-end of life situation. However, and as acknowledged by Pennington *et al.* (2015), willingness-to-pay valuations made by individuals facing the prospect of imminent death can be expected to be high because the opportunity costs in those circumstances are low or non-existent. Other than the ability to leave

a legacy, money is arguably of no use to individuals when they are dead. This is often referred to as the 'dead-anyway' effect whereby an increase in an individual's mortality risk reduces their expected marginal utility of wealth (thereby increasing their willingness-to-pay) since the marginal utility of wealth when alive is greater than the marginal utility of wealth when dead (Pratt and Zeckhauser, 1996). It is therefore understandable and perhaps consistent with utility-maximising behaviour for individuals nearing their end of life to be willing to spend most or all of the money they have on extending their life, even if the utility gains from the life extension are small. If such willingness-to-pay values are then used to inform decisions about how to spend a common pool of funding that has been raised from members of the public (many of whom will not be at their end of life), then the opportunity cost of expenditure on end of life treatments will be higher as it would result in foregoing spending on other treatments. It may therefore be considered inappropriate to use willingness-to-pay values elicited from an individual perspective to inform society-level decision-making. It should be noted, however, that Shiroiwa *et al.* (2010) observed higher values for gains accruing to respondents' family members and to unidentified members of society than those accruing to the respondents themselves in five of the six countries studied. The authors suggest that this result may reflect altruistic preferences.

When developing the inclusion criteria for this review, it was deemed appropriate to include own health perspective studies that clearly sought to inform health care priority-setting policies. Some own health perspective studies that appeared to report results of potential relevance to the overall research question were nevertheless excluded on the basis that they did *not* clearly seek to inform health care priority-setting policies (e.g. Kvamme *et al.*, 2010). An alternative approach would have been to restrict the review to studies adopting a social decision-maker perspective. One of the studies that used the willingness-to-pay method would continue to be included in the review on the basis that it also reported preferences obtained using person trade-off tasks undertaken from a social decision-maker perspective (Pinto-Prades *et al.*, 2014). The study by Shiroiwa *et al.* (2010) would be excluded on the basis that it employed a social decision-maker perspective in only one task, involving a scenario describing imminent death, so comparisons between end of life and non-end of life social decision-maker valuations would not be possible. Applying such a restriction would result in a slightly different balance of findings across the studies: of the studies that would remain, five report evidence consistent with an end of life premium and eight do not.

2.4.2 Inclusion of indifference options

Studies that offered respondents the opportunity to express indifference between the alternatives on offer were more likely to report evidence consistent with an end of life premium than those that did not. The nature of the indifference options available differed across studies. In the choice exercise studies, options such as 'Can't decide' (Stahl *et al.*, 2008) and 'I have no preference' (Shah *et al.*, 2014) were presented. In the willingness-to-pay studies, respondents could express indifference by stating the same value for two or more different gains. In the budget allocation study, respondents could choose to split resources evenly between the two recipient groups. In the Q methodology study, respondents were required to position seven of the 49 statements in such a way that implied neither agreement nor disagreement.

The way in which indifference options are framed can affect respondents' willingness to choose those options – for example, Shah and Devlin (2012) reported that respondents showed an attraction to a 50:50 split when asked to allocate a budget between two patient groups but an aversion to an 'I have no preference' option in a choice exercise involving the same two groups. This finding is supported by those of the present review – the sole budget allocation study found that a 50:50 split was the most popular option, and consequently was one of only three studies that both offered an indifference option and did not report evidence consistent with an end of life premium. It may be that respondents consider a 50:50 split (but not an 'I have no preference' response) to be a legitimate choice when they find it difficult to choose between two options. Alternatively, they may be concerned about the implications of expressing indifference in a choice exercise – for example, they might be under the impression that failing to choose means that neither patient would receive the treatment on offer.

When respondents are indifferent between the available options but no indifference option is available, they are forced to make a choice in order to proceed. In principle, these respondents should make their choices at random, which will tend to result in a roughly even split between the available options in the choice data. In practice, respondents may pursue an alternative choice strategy. For example, when faced with a choice between treating an end of life patient and a non-end of life patient, a respondent may anticipate other respondents choosing to treat the end of life patient but may themselves consider both patients to be equally deserving of treatment. If this respondent wishes that both patients should be given an equal opportunity to be treated, they may then express a preference for treating the non-end of life patient (to counteract the choices they anticipate the

other respondents making). This increases the likelihood of the study failing to find an overall preference for treating the end of patient. Evidence of such response behaviour has been discussed by Shah *et al.* (2015b).

It is common for DCEs and studies using internet surveys – both of which are becoming increasingly popular in this field – not to include opt-out or indifference options. For DCEs, best practice guidelines advise that indifference options are often inappropriate as they can have implications for the experimental design and lead to the censoring of data (Bridges *et al.*, 2011). For internet surveys, which are sometimes viewed with suspicion due to concerns about respondents' attentiveness, indifference options are often avoided on the grounds that they will be used a default choice, thus providing respondents with a way to avoid taking time to make difficult decisions. If studies are less likely to detect support for an end of life premium if they do not include an indifference option, and if the trend for studies not to include an indifference option continues, then it can be expected that fewer studies will report evidence consistent with an end of life premium going forward.

2.4.3 Use of visual aids

The use of visual aids appears to be increasing. All but one of the seven studies published since 2014 included diagrams designed to help respondents make sense of the (often complex) choice tasks. These often took the form of figures depicting quality of life on one axis and length of life or time on the other. Visual aids were used in all five DCE studies reviewed, and in the majority of studies administered using a computer-based approach.

Studies that used visual aids were more likely to report evidence consistent with an end of life premium than those that did not. One possible explanation is that very short amounts of time (in most studies respondents were presented with scenarios in which at least one patient had less than 12 months left to live) appear starker and more dramatic when presented graphically than when described verbally.

It has been argued that graphs may not be the best way to present scenario information to survey respondents due to concerns that they unintentionally lead to different respondents interpreting the information in different ways (van de Wetering *et al.*, 2015). For example, when faced with diagrams in which better quality of life and longer life expectancies are represented by larger areas, some respondents may (subconsciously or otherwise) be attracted to the larger areas and therefore to the alternatives depicted by diagrams showing longer life expectancies.

Such framing effects are clearly a matter of concern, particularly in studies where no interviewer is present, since the opportunities for instructing and debriefing respondents are very limited. This makes it difficult to know for certain the extent to which the choice data truly reflect the respondents' beliefs and preferences, or whether the respondents interpreted and answered the questions as the researcher had intended them to. However, this concern is not restricted to the use of visual aids. One possibility is that respondents being presented with two or more hypothetical patients may mistakenly interpret the task as asking them which patient they would prefer to be in the position of rather than which patient they consider to be more deserving of treatment. It is not clear that such a misinterpretation would be more likely to occur in a survey using a combination of text and graphical descriptions than in one using only text descriptions. Indeed, if the issue is that respondents being presented with complex choice tasks do not always understand what is being asked of them, it seems intuitive to give them more, rather than less, assistance.

Further, if the use of visual aids encourages respondents either to choose the patient they would prefer to be in the position of, or to choose the alternative associated with larger areas, then this would in most cases result in them being more likely to choose to treat patients with longer rather than shorter life expectancies. This is inconsistent with the finding of this review that studies using visual aids were more likely to report evidence consistent with an end of life premium than those that did not. One study that used two different question frames to understand respondents' preferences regarding end of life found that many respondents expressed support for prioritising life-extending end of life treatments in the DCE tasks (which used visual aids) (Rowen *et al.*, 2016a). However, the same respondents then gave responses to more direct attitudinal questions (which did not use visual aids) that suggest that they did not believe that the NHS should give priority to such treatments. Furthermore, the one study that actively set out to examine the impact of visual aids found that the propensity to choose to treat the patient group with shorter life expectancy was unaffected by whether or not diagrams were used to illustrate the information (Rowen *et al.*, 2016b).

The findings of this review may suggest that the likelihood of a study providing evidence consistent with an end of life premium is linked to the choice of elicitation preference method and to whether indifference options and visual aids were used. However, it should also be noted that conflicting results were reported by two studies that did not differ in these respects. Shah *et al.* (2015a) and Rowen *et al.* (2016a) both used the DCE method with forced-choice tasks supported by visual

aids (indeed, Shah *et al.* acknowledge that they based their design on that of the Rowen *et al.* study, using very similar graphs and text descriptions to present information to respondents). Both studies also used similar samples – members of the UK public recruited from online panels and broadly representative of the general population in terms of age and gender.

2.4.4 Limitations

Some limitations of the review should be mentioned. Only one database – the SSCI – was searched. This is an interdisciplinary database covering around 3,000 journals, including most major health economics and health policy journals known to the author. However, its focus is on social sciences, and does not cover all specialist medical and scientific journals that may have published articles that are relevant to the research question.

The approach to identifying terms for the electronic search involved analysing the abstracts of key papers already known to the author to detect unique words that appeared in at least two of those abstracts. This 'searching for search terms' exercise was useful and led to the selection of additional terms that increased the specificity of the search. However, it could have been extended by examining the titles and keywords of the key papers in addition to their abstracts. For example, terms such as *priority* and *empirical* appear in the titles of the key papers but not in their abstracts, and were therefore overlooked.

Throughout the search strategy development process, efforts were made to make the search more efficient – that is, to reduce the number of hits without losing any relevant records, and to increase the number of relevant records without substantially increasing the number of hits. The impact of changes to the search strategy was tested by informally assessing the results each time a change was made. For example, when adding a search term a selection of additional records identified was examined – the titles and abstracts of the most recent 20 records were scanned. A more robust approach would have been to *randomly* select the sample of 20 records. By default, the search results were ordered by publication date, so looking only at the first few records rather than at a random selection will have led to bias if recently published articles tend to use different terminology from older articles.

The review included only articles that have been published in English. Only two records were excluded due to publication in a language other than English (Figure 2-1), but this could be linked to the choices made regarding data sources and the search strategy. It is acknowledged that there may be studies that are relevant to the research question that have been published in other languages, and alternative

data sources could have been examined in order to identify these. However, it would have been very difficult to review these records, given the time constraints and limited foreign language skills of the author.

On a related note, the review was to a large extent motivated by the policy context in the UK. The author of this review identifies as a health economics researcher based in the UK (the same is true of many of the authors of studies included in the review). Hence, the search terms considered are likely to reflect the language used by this particular subset of the academic community and may not be well suited for identifying, say, articles authored by ethicists or by researchers based in low and/or middle income countries.

Only articles that elicited stated preferences with the intention of informing health care priority-setting policies were considered. There were a large number of studies that may have contained informative data but did not relate to a priority-setting or resource allocation context, such as surveys of end of life patients' own preferences for living longer or maintaining quality of life. However, these studies were not relevant to the research question and therefore did not meet the criteria for inclusion in the review.

Whereas reviews of clinical trials are subject to rigorous guidance on search methods, data extraction and evidence synthesis (Centre for Reviews and Dissemination, 2009; Liberati *et al.*, 2009), such guidance is unavailable for reviews of stated preference studies. Although it cannot be claimed that the review is fully exhaustive, efforts have been made to be explicit about the methods used (and the reasons for using them) and balanced in the presentation of findings. Some of the limitations and potential biases associated with the approach have been acknowledged, and efforts have been made to minimise these as far as possible.

As mentioned above, a formal assessment of study quality was not undertaken due to the lack of a known, standardised method for doing so. Instead, publication in a peer-reviewed journal was relied on as a proxy for quality. None of the studies included in the review was judged to be of such poor quality that their findings ought to be disregarded. However, it is acknowledged that there may be studies that are relevant to the research question that have not been published in a peer-reviewed journal, such as those in the grey literature (for example, reports of NICE's Citizens' Council – see NICE, 2017a) and working papers or theses that have never been submitted to or accepted by a journal.

The follow-up of reference lists of the articles whose full texts were reviewed was useful – eight of the 20 included articles (40%) were identified in this way. A

further step would have been to search for articles that have cited those already identified. This technique forms part of a search method known as 'snowballing' or 'citation pearl growing' (Paisley, 2014).

2.4.5 Gaps in the literature

Given the possibility that the findings of stated preference studies are influenced by the choice of elicitation method or by characteristics of the study design, it would be informative for studies to use multiple methods or designs in order to test the robustness of their results. Most of the studies included in this review used a single method and design throughout. Exceptions to this include Pinto-Prades *et al.* (2014), who noted discrepancies between willingness-to-pay and person trade-off responses at the within-respondent level (though the same broad conclusion was reached using both methods); and Rowen *et al.* (2016b), who compared the results achieved using different modes of administration and question framings.

A related issue is that few studies sought to understand whether respondents would agree with the researchers' interpretations of their responses to the stated preference tasks. Rowen *et al.* (2016a) inferred from their DCE data that there was robust and consistent support for an end of life premium. Yet when asked about the prioritisation of end of life patients more directly later in the survey, the majority of respondents expressed views that implied the opposite conclusion. It would be informative for researchers to test the stability of respondents' preferences – for example, by presenting the policy implications of their earlier choices and checking whether they agree with these (Whitty *et al.*, 2014; Shah *et al.*, 2015b). Studies applying techniques that are designed to allow unexpected views to emerge, such as Q methodology, also offer promise for researchers seeking to make sense of apparently inconsistent or counterintuitive preferences.

As described above, an observed preference for giving priority to the treatment of end of life patients may in fact be driven by age- or time-related preferences. Several studies in this review did not control for (or even mention) age-related preferences, and only one study attempted to control for time-related preferences. If age- and time-related preferences are not controlled for, it will not always be clear what exactly is driving any observed preference for treating patients with shorter life expectancy, particularly if the differences between the life expectancies of the candidate beneficiaries are relatively large (e.g. Dolan and Cookson, 2000). In the only study that attempted to control for time-related preferences (Shah *et al.*, 2014), the authors conjecture that the choices made by respondents may have been driven by concern about the suddenness of the patients' disease progression, and therefore how much time they have had to 'prepare for death', rather than the

fact that they are at the end of life *per se*. Preferences regarding preparedness have received limited attention in the literature to date, and further investigation of this issue would be welcomed.

2.5 Conclusions

The literature review presented in this chapter has shown that the evidence on public preferences regarding end of life treatments is limited (but growing) and mixed. The research reported in the remainder of this thesis contributes to the evidence base – indeed, empirical studies 2 and 3 (Chapters 4 and 5, respectively) were included in the review, having been published in peer-reviewed journals prior to May 2016. All four empirical studies in this thesis attempt – using a variety of samples, methods and modes of administration – to shed light on the overarching research 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.

As noted in 1.9, the evidence was particularly scarce at the time when NICE's policy was introduced. None of the studies included in the review that were published prior to 2013 explicitly set out to examine end of life-related preferences. Empirical studies 1 and 2 – both of which were conducted in 2011 – were amongst the first to investigate public preferences with specific regard to the NICE criteria. Empirical study 3 was conducted in 2012, at which time there was a dearth of large-scale studies of preferences of the UK public – the UK studies that had been published at the time had mostly used small samples of 100 respondents or fewer. It was also one of the first studies to have applied the DCE method in the end of life context.

The review has highlighted a number of potential trends and gaps in the existing literature (as of May 2016). It is hypothesised that the findings of a given study of end of life-related preferences will be influenced by design choices such as the perspective respondents are asked to adopt, whether an indifference option is available, and whether visual aids are used to present information. Empirical study 4 (Chapter 6) uses a multi-arm design to examine the impact of these factors. It also attempts to test the stability of respondents' preferences by using two different types of question to elicit information on support for an end of life premium.

3 VALUING HEALTH AT THE END OF LIFE: AN EXPLORATORY PREFERENCE ELICITATION STUDY (EMPIRICAL STUDY 1)[15]

3.1 Introduction

This chapter describes an exploratory preference elicitation study conducted in early 2011. It is the first of four empirical studies reported in this thesis, and can be considered a pilot for empirical study 2 (Chapter 4). The aim of the study is to design and pilot an approach to examining whether a policy of giving higher priority to life-extending end of life treatments (as specified by NICE) than to other types of treatments is consistent with the preferences of members of the public.

At the time the study was conducted, it had been acknowledged that there was little evidence of public support for the premise underpinning NICE's end of life policy. No comparable studies were available – the studies reviewed in Chapter 2 that had been published prior to 2011 had all set out to address different research questions.

3.2 Methods

3.2.1 Survey instrument

A survey was developed in which respondents were presented with choice tasks based on five scenarios (S1 to S5). All respondents considered all five scenarios, in the same order. Each scenario presented respondents with information about two hypothetical individuals (patient A and patient B) who have been diagnosed with illness. This information was presented using written descriptions and diagrams. Both patients could benefit from treatment but the respondents were asked to assume that the health service had enough funds to treat one but not both of them. The question posed to respondents was: "Would you prefer to treat patient A or patient B?" (though they were also permitted to indicate that they had no preference; see below). The scenarios are replicated in full in Appendix 4, and summarised in Table 3-1.

In scenarios S1, S2 and S3, treatment would extend the life of either patient A or patient B by six months (with certainty). The patients differed in terms of age and the amount of time between diagnosis and expected death. These scenarios did not

[15] A previous version of this chapter has been published as: Shah, K., Wailoo, A. and Tsuchiya, A., 2011. *Valuing health at the end of life: an exploratory preference elicitation study*. OHE Research Paper. London: Office of Health Economics.

examine quality of life – respondents were advised that the patients' illnesses were asymptomatic and that treatment would not affect their quality of life.

In S4 and S5, the illnesses were described as having a negative effect on quality of life, with both patients experiencing their final year of life at 50% of quality of life (described to respondents as 'full health'). In these scenarios, treatment would restore patient A to full health (with no effect on life expectancy) or extend the life of patient B by one year (with no effect on quality of life). In all cases, the patients' prognoses and gains from treatment were described as if they were known with certainty.

Under the conditions that two years in 50% health is equivalent to one year in full health, and that a health gain today is equivalent to an equal-sized health gain in the future, both patient A and patient B will gain exactly the same amount of health from treatment in all five scenarios – half of a QALY. However, no specific explanation of the meaning of 50% health was provided to respondents.

Table 3-1. Summary of scenarios used in the empirical study 1

	S1	S2	S3	S4	S5
Without treatment					
Patient A life expectancy	10 years	1 year	10 years	1 year	10 years
Patient A quality of life	100%	100%	100%	50%	50%
Patient B life expectancy	1 year	1 year	1 year	1 year	1 year
Patient B quality of life	100%	100%	100%	50%	50%
Gains from treatment					
Patient A life expectancy	+6 mths	+6 mths	+6 mths	No change	No change
Patient A quality of life	No change	No change	No change	+50%	+50% [a]
Patient B life expectancy	+6 mths	+6 mths	+6 mths	+1 year	+1 year
Patient B quality of life	No change	No change	No change	No change	No change
Undiscounted QALY gain from treatment [b]					
Patient A	0.5 QALY	0.5 QALY	0.5 QALY	0.5 QALY	0.5 QALY
Patient B	0.5 QALY	0.5 QALY	0.5 QALY	0.5 QALY	0.5 QALY
Age of patients	A & B are same age	A & B are same age	B is 9 years older than A	A & B are same age	B is 9 years older than A
Timing of scenario (when does the treatment decision occur)	At time of A & B's diagnosis	9 years after A's diagnosis	At time of A & B's diagnosis	At time of A & B's diagnosis	At time of A & B's diagnosis

[a] Quality of life gain in final year of life only
[b] Respondents did not see this information (the term 'QALY' was not used at any point in the survey)

Scenario S1 provides a simple test of whether respondents wish to give higher priority to the treatment of end of life patients, as the only difference between the two patients at the start of the scenario is that patient B has a shorter amount of time left to live than patient A (one year would be classed as 'short life expectancy' under the first criterion of NICE's end of life policy, whereas 10 years would not).

However, the scenario design is such that there may be reasons other than favouring the treatment of end of life patients for choosing to treat patient B in S1. First, without treatment patient A will be nine years older when they die than

patient B will be when they die. Hence, a preference to treat patient B may be driven by a social preference for giving priority to the young. There is some evidence in the literature of public support for age weighting based on equity concerns (Dolan *et al.*, 2005; Whitty *et al.*, 2014; Gu *et al.*, 2015). To address this issue, scenario S3 replicates S1 except that patient B is nine years older than patient A at the start of the scenario, which means that both patients will die at the same age without treatment.

Second, the benefit from treating patient A would not take place until 10 years into the future (compared to one year into the future for patient B). Hence, a preference for treating patient B may be driven by a preference for enjoying benefits sooner rather than later. In general, it is assumed by health care decision-makers in the UK that society has a 'positive time preference' (HM Treasury, 2003), which means that the further into the future benefits are accrued, the lower the value of those benefits. To address this issue, scenario S2 replicates S3 except that patient A was diagnosed with their illness nine years prior to the start of the scenario. This means that the benefits from treating patient A would now take place one year into the future – the same as for patient B. Thus both patients are at the 'end of life' in S2, but patient B has progressed to this stage more suddenly than has patient A.

As mentioned above, considerations of quality of life are introduced in scenarios S4 and S5. S4 involves choosing between treatments that extend life and treatments that improve quality of life. NICE's end of life criteria accommodate life extensions but not quality of life improvements. Scenario S5 combines elements of S3 and S4 in that it involves choosing between treating a non-end of life patient (patient A) and an end of life patient (patient B) and between a quality of life-improving treatment (to patient A) and a life-extending treatment (to patient B).

Conjectured explanations for different responses to selected key combinations of scenarios are presented in Table 3-2. Note that 'time preference' refers to a preference for enjoying benefits sooner rather than later; 'age preference' refers to a preference for favouring the treatment of younger patients; and 'end of life preference' refers to a preference for favouring the treatment of patients with short life expectancy who have little time to prepare for death due to the suddenness of disease onset. It is conjectured that a supporter of NICE's end of life policy would choose to treat patient B in all scenarios, except perhaps S2 where it is unclear whether the supplementary advice applies (the criteria do not distinguish between sudden and non-sudden onset or progression of disease).

Table 3-2. Conjectured explanations for responses to selected scenario combinations

Scenario	Choice	Scenario	Choice	Conjectured explanation
S1	A	S2	A	Preferences run counter to end of life criteria
S1	A	S2	B	Reverse time and/or age preference
S1	A	S2	I	
S1	I	S2	B	
S1	B	S2	A	Time and/or age preference
S1	B	S2	I	
S1	I	S2	A	
S1	B	S3	B	End of life preference
S1	A	S3	A	Preferences run counter to end of life criteria
S1	A	S3	B	Reverse age preference
S1	A	S3	I	
S1	I	S3	B	
S1	B	S3	A	Age preference
S1	B	S3	I	
S1	I	S3	A	
S1	B	S3	B	End of life preference and/or time preference
S2	A	S3	A	Preferences run counter to end of life criteria
S2	A	S3	B	Time preference
S2	A	S3	I	
S2	I	S3	B	
S2	B	S3	A	Reverse time preference
S2	B	S3	I	
S2	I	S3	A	
S2	B	S3	B	End of life preference
S4	A	S5	A	Preference for quality of life-improving treatments
S4	A	S5	B	End of life preference and/or time preference
S4	A	S5	I	
S4	I	S5	B	
S4	B	S5	A	Preferences run counter to end of life criteria / reverse time preference
S4	B	S5	I	
S4	I	S5	A	
S4	B	S5	B	Preference for life-extending treatments
S3	A	S5	A	Preferences run counter to end of life criteria
S3	A	S5	B	Preference for life-extending treatments
S3	A	S5	I	
S3	I	S5	B	
S3	B	S5	A	Preference for quality of life-improving treatments
S3	B	S5	I	
S3	I	S5	A	
S3	B	S5	B	End of life preference and/or time preference

A = respondent prefers to treat patient A; I = respondent indicates that they have no preference between treating patient A and treating patient B; B = respondent prefers to treat patient B

3.2.2 Administration of survey

The survey was administered using face-to-face interviews undertaken by the author. All interviews were carried out in a one-to-one setting in meeting rooms at the University of Sheffield's School of Health and Related Research.

Respondents were given a paper questionnaire (Appendix 4) and considered the scenarios one at a time. They were asked to read the description for each scenario before informing the interviewer of their answer. The aim was to elicit considered

responses, so respondents were encouraged to 'think aloud' and to discuss the reasons for their choices with the interviewer. They were also permitted to amend their responses in earlier scenarios if they changed their mind during the course of the interview. The interviewer emphasised that a 'no preference' response was acceptable.

After completing the final scenario, the respondents were asked a series of probing questions designed to elicit qualitative information about the thinking behind their responses. Background information (age, gender, experience of serious illness) was collected at the end of the interview. All interviews were audio recorded with the permission of the respondents.

3.2.3 Sample

The survey was administered on a convenience sample of members of non-academic staff and postgraduate research students at the University of Sheffield (excluding those in the Faculty of Medicine, Dentistry and Health). The target sample size of 20 respondents was determined on the basis of available time and resources. The sample was recruited using two methods: (1) email invitation to participate sent to administrative, facilities, specialist and technical staff and postgraduate research students at the University of Sheffield (excluding those in the Faculty of Medicine, Dentistry and Health); and (2) recruitment flyer posted in areas used by facilities staff at the University of Sheffield. Respondents received a £5 gift voucher to thank them for their participation.

3.2.4 Ethical approval

The survey design 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.

3.2.5 Methods of analysis

Descriptive statistics were reported in order to summarise the sample and responses to the scenario questions. For the pairs of scenarios presented in Table 3-2, the numbers and proportions of respondents making each combination of choices were analysed using cross-tabulations. Due to the small sample size and exploratory nature of the study, no statistical tests were conducted.

Comments made by respondents, either when explaining their choices or when responding to the probing questions, were summarised by the author using paraphrasing or direct quotations.

3.3 Results

Interviews were completed by 21 respondents in April 2011. The background characteristics of the sample are presented in Table 3-3. The sample includes a larger proportion of females than in the general population (Office for National Statistics, 2011). The age distribution is broadly similar to that of the general population when only individuals of working age (18 to 65 years) are considered. The convenience nature of the sample means that it excludes individuals who are unemployed and those who are employed in a professional role.

Table 3-3. Sample background characteristics

		n	%	Population
Total		21	100.0%	
Age (years)	18-29	4	19.0%	21%
	30-44	7	33.3%	26%
	45-59	9	42.9%	25%
	60+	1	4.8%	28%
Gender	Female	15	71.4%	51%
	Male	6	28.6%	49%
Experience of serious illness	In themselves	1	4.8%	
	In their family	17	81.0%	
	In caring for others	7	33.3%	
Work status [a]	Staff	20	95.2%	
	Student	2	9.5%	

[a] One respondent was included under both categories (part-time postgraduate student, part-time administrative staff)

All interviews lasted for between 20 and 35 minutes. All 21 respondents completed all five scenarios and answered all of the relevant probing questions. Data saturation was reached after approximately 15 interviews.

3.3.1 Response data

Table 3-4 reports aggregate response data for each of the five scenarios. Four respondents (19.0%) preferred to treat patient B in all five scenarios.

Table 3-4. Aggregate response data for all scenarios

	S1	S2	S3	S4	S5
Prefer to treat patient A	3 (14.3%)	0 (0.0%)	6 (28.6%)	12 (57.1%)	6 (28.6%)
No preference	2 (9.5%)	4 (19.0%)	3 (14.3%)	3 (14.3%)	2 (9.5%)
Prefer to treat patient B	16 (76.2%)	17 (81.0%)	12 (57.1%)	6 (28.6%)	13 (61.9%)
Total	21 (100.0%)	21 (100.0%)	21 (100.0%)	21 (100.0%)	21 (100.0%)

Table 3-5 to Table 3-9 provide cross-tabulations of the response data for the combinations of scenarios shown in Table 3-2. In these tables, the value in each cell refers to the number of respondents expressing that set of preferences.

Table 3-5. Cross-tabulation – S1 versus S2

	S2			
S1	Prefer A	No preference	Prefer B	Total
Prefer A	0 (0.0%)	0 (0.0%)	3 (14.3%)	3 (14.3%)
No preference	0 (0.0%	2 (9.5%)	0 (0.0%)	2 (9.5%)
Prefer B	0 (0.0%)	2 (9.5%)	14 (66.7%)	16 (76.2%)
Total	0 (0.0%)	4 (19.0%)	17 (81.0%)	21 (100.0%)

In S1, patient A and patient B are the same age today, and without treatment patient A will be nine years older when they die than patient B will be when they die; the treatment decision occurs at the time of diagnosis for both patient A and patient B. In S2, patient A and patient B are the same age today, and without treatment both patients will be the same age when they die; the treatment decision occurs at the time of diagnosis for patient B and nine years after the time of diagnosis for patient A.

Most respondents preferred to treat patient B in both scenarios S1 and S2. In general, they claimed (both whilst thinking aloud and when answering the probing follow-up questions) that the rationale behind their choices was the same in both scenarios, but that the choice in S2 was more difficult.

Three respondents preferred to treat patient A in S1. The main argument given for this was that it was 'not worth' giving an extra six months to someone with as short a time to live as patient B – rather, the life extension would be more valuable if given to someone who has more time to participate in society and who has a better opportunity to get their life in order (and could therefore make the most out of the additional time offered by treatment). All three respondents then switched to choosing to treat patient B in S2. Despite interviewer probing, the respondents were unable to articulate clearly the reasons for this shift in choices.

Two respondents switched from choosing to treat patient B in S1 to having no preference in S2, arguing that although patient A would have had more time to prepare for death than patient B in S2, it would be unfair to assume that they will have made good use of that time.

Table 3-6. Cross-tabulation – S1 versus S3

	S3			
S1	Prefer A	No preference	Prefer B	Total
Prefer A	3 (14.3%)	0 (0.0%)	0 (0.0%)	3 (14.3%)
No preference	0 (0.0%)	2 (9.5%)	0 (0.0%)	2 (9.5%)
Prefer B	3 (14.3%)	1 (4.8%)	12 (57.1%)	16 (76.2%)
Total	6 (28.6%)	3 (14.3%)	12 (57.1%)	21 (100.0%)

In S1, patient A and patient B are the same age today, so without treatment patient A will be nine years older when they die than patient B will be when they die. In S3, patient B is nine years older than patient A, so without treatment both patients will be the same age when they die.

Of the respondents who preferred to treat patient B in S1, the majority also preferred to treat patient B in S3, although one switched to having no preference and three switched to choosing to treat patient A. The respondent who switched to having no preference expressed worry about the presence of specific information on patients' ages – whilst thinking aloud, they said to themselves: "is it justifiable to take these things into account?" They eventually said that they were unwilling to choose between the patients because they did not feel that it was appropriate to make prioritisation decisions based on current age. They added that if they were forced to choose then then could not help but take age into account, given the prominence of the information about age in the scenario description. They chose not to amend their responses to the previous scenarios.

Of the three respondents who switched to preferring to treat patient A in S3, two said that their decision was based on a concern for treating the young ("gives the chance to the younger patient"). The fact that both patients would die at the same age without treatment was not considered important by these respondents – their concern was about how old the patients are *now*. The third respondent who made this switch said that they had imagined that the patients were children, and that their preference for treating patient A was based on a desire or duty to protect the very young.

It should be noted that although most respondents quickly recognised that the only difference between S1 and S3 was the ages of the patients, the two that did not immediately recognise this both switched from preferring to treat patient B in S1 to preferring to treat patient A in S3. However, both respondents displayed a good level of understanding after clarification had been provided by the interviewer, so their choices should not be interpreted as having been driven by misunderstanding.

Table 3-7. Cross-tabulation – S2 versus S3

	S3			
S2	Prefer A	No preference	Prefer B	Total
Prefer A	0 (0.0%)	0 (0.0%)	0 (0.0%)	0 (0.0%)
No preference	1 (4.8%)	2 (9.5%)	1 (4.8%)	4 (19.0%)
Prefer B	5 (23.8%)	1 (4.8%)	11 (52.4%)	17 (81.0%)
Total	6 (28.6%)	3 (14.3%)	12 (57.1%)	21 (100.0%)

In S2, the treatment decision occurs at the time of diagnosis for patient B and nine years after the time of diagnosis for patient A. In S3, the treatment decision occurs at the time of diagnosis for both patient A and patient B.

Of the respondents who preferred to treat patient B in S2, 64.7% preferred to treat patient B in S3; the majority of the remaining respondents switched to preferring to treat patient A. One respondent chose to treat patient B in S3 but had no preference in S2. Their reasoning was that patient A could be viewed as the better off of the two patients in S3 (and indeed in S1) due to having nine extra years to

prepare for death, but it seems unfair to take this into account in S2 as patient A may not have made good use of this time. Five respondents went further, choosing to treat patient A in S3. Some of these respondents did so due to a concern for the young (described above in the discussion of S1 versus S3), whilst the others did so due to a belief that the life extension should be given to the patient who has more time to participate in society (described above in the discussion of S1 versus S2).

Table 3-8. Cross-tabulation – S4 versus S5

	S5			
S4	Prefer A	No preference	Prefer B	Total
Prefer A	6 (28.6%)	1 (4.8%)	5 (23.8%)	12 (57.1%)
No preference	0 (0.0%)	1 (4.8%)	2 (9.5%)	3 (14.3%)
Prefer B	0 (0.0%)	0 (0.0%)	6 (28.6%)	6 (28.6%)
Total	6 (28.6%)	2 (9.5%)	13 (61.9%)	21 (100.0%)

In both S4 and S5, treating patient A would generate a quality of life improvement and treating patient B would generate a life extension. In S4, without treatment both patient A and patient B will live for one year in 50% health. In S5, without treatment patient A will live for 10 years in 50% health and patient B will live for one year in 50% health.

Just over half of the respondents preferred to treat patient A in S4, which indicates that they prefer to give priority to quality of life-improving rather than life-extending treatments for patients with one year of life expectancy. Half of those respondents then switched either to preferring to treat patient B or to having no preference in S5. A conjectured explanation for these combinations of choices is that whilst a preference for quality of life-improving treatments continues to exist in S5, this preference is outweighed by the preference to prioritise the treatment of those with short life expectancy. In general, the comments made by respondents support this explanation.

Table 3-9. Cross-tabulation – S3 versus S5

	S5			
S3	Prefer A	No preference	Prefer B	Total
Prefer A	4 (19.0%)	0 (0.0%)	2 (9.5%)	6 (28.6%)
No preference	0 (0.0%)	1 (4.8%)	2 (9.5%)	3 (14.3%)
Prefer B	2 (9.5%)	1 (4.8%)	9 (42.9%)	12 (57.1%)
Total	6 (28.6%)	2 (9.5%)	13 (61.9%)	21 (100.0%)

Of the 12 respondents who chose to treat patient B (the end of life patient) in S3, most also chose to treat patient B in S5. Three respondents switched to preferring to treat patient A or having no preference in S5 – all three claimed that the preference for quality of life-improving treatments outweighed (or in one case, cancelled out) the preference for prioritising end of life treatments in this scenario.

3.3.2 Responses to probing questions

Responses to the probing questions are summarised below.

Q1. How did you find the survey?

Almost all of the respondents said that they found the survey interesting. Some mentioned that the scenarios were unpleasant to think about, but none suggested that they regretted taking part. This supports the interviewer's observation that all respondents were highly engaged throughout the interviews.

Q2. To what extent do you feel you understood the questions being asked?

Almost all of the respondents said that they felt that they had understood the questions well, although in a few cases this was not always consistent with the interviewer's observation that they were hesitant or confused at some points during the interview. Where misunderstanding did occur, respondents typically blamed their inattentiveness rather than the way in which the information had been presented.

Q3. What did you think about the graphical illustrations of the scenarios?

Most of the respondents said that the diagrams were helpful. A few respondents were particularly approving, claiming that they relied on them heavily. On the other hand, some respondents said that they did not use them at all. A few respondents indicated that they were confused by the distinction between health without treatment and health gains from treatment in the diagrams.

Q4. How difficult did you find it to decide on your answers?

Some respondents said that they found it very difficult to decide without more information (in particular, information on whether the patients were children or adults); others said that they found it easier to make decisions without such complicating factors.

The general consensus was that scenarios S4 and S5 were more difficult to answer than the previous three scenarios. This was due in part to respondents not having clear or considered reasons for preferring either quality of life improvements or life extensions, and in part to the lack of clarity about what exactly is meant by '50% health'.

Q5. In some of the tasks, you preferred to treat neither patient A nor patient B. Can you tell me a bit more about why you were unwilling or unable to choose between them?

Only six respondents expressed 'no preference' in any of the scenarios. One respondent declined to choose between patient A and patient B in all scenarios. They felt that both patients were equally entitled to treatment, and that differences

between the patients are not relevant in terms of their claims to health care. Other reasons given for expressing no preference included: not having enough information to justify choosing between the two patients; belief that the patients are equally worthy of treatment; and lack of clarity about what is meant by '50% health' (one respondent claimed that if 50% health still allows you to 'enjoy what's left', then they would prefer to treat patient B in S4, but if 50% health would prevent you from enjoying your remaining life, then they would prefer to treat patient A).

Q6. When deciding which patient to treat, what sorts of things did you take into account?

Aside from the factors discussed in probing questions 7 to 10 (see below), a number of other considerations were mentioned in response to this question. Several respondents said their thinking had been guided by personal experiences of seeing friends and family in serious ill health. Several respondents spoke of trying to put themselves 'in the shoes' of the hypothetical patients – that is, trying to imagine what they would want for themselves if they were in the position of patient A or patient B.

A number of respondents referred to the idea of treatment giving patient B a greater 'proportional' or 'percentage' gain in life extension. In S1, for example, these respondents claimed that their preference for treating patient B would hold as long as patient B was gaining proportionately more time than patient A (i.e. more than 2.6 weeks). Some respondents referred to an objective of achieving fairness and/or equality, suggesting that the treatment should be given to whichever patient is deemed to be the 'worse off'.

Regarding scenarios S4 and S5, some respondents justified their preference for treating patient A by explaining that whereas they could reasonably assume that 'everyone wants better quality of life', it could not be assumed that 'everyone wants to live longer'.

Other considerations mentioned included: 'suffering'; 'how much one treasures life'; the personal and family circumstances of the patients; how able patients are to adapt to the idea that they are dying; and any other health problems that the patients might be facing.

Q7. When deciding which patient to treat, did you think about how old the patients would be when they die?

Most respondents said that they thought about age but did not take it into account when making their choices. As described above, those respondents that *did*

explicitly take age into account tended to be more concerned about age at treatment than about age at death. Some respondents felt strongly that age should not be used as a priority-setting criterion.

Q8. When deciding which patient to treat, did you think about whether the patients would have sufficient time to 'prepare for death'?

Almost all of the respondents said that they took this factor into account. Of those that did not, one respondent questioned whether having 10 years to live before a specified time of death is a good or a bad thing (due to disutility from knowing that you are going to die). Another respondent said that they were "not from a culture that does great death preparation" and therefore questioned the value of having extra 'time to prepare'.

Q9. When deciding which patient to treat did you think about the fact that some of the benefits of treatment wouldn't take place until far away into the future?

Almost all of the respondents answered no to this question, claiming that the only way in which timing matters is in terms of how much time the patients have in good health before reaching their end of life. A few respondents said that when considering longer timeframes it would be reasonable to hope for the possibility of medical breakthroughs or further treatment (if the respondents brought this up whilst considering the scenarios, the interviewer emphasised that it should be assumed that further treatment would *not* be possible).

Q10. Thinking about scenarios 4 and 5, when deciding which patient to treat, did you think about the possibility that being in only 50% health would involve being a burden on others or on society?

A slight majority of respondents said that they did not think about this possibility. Some respondents said that they were aware of but specifically ignored this issue as they were not asked to consider it. A few respondents rejected the idea that people in poor health can be described as a burden. Of those that considered this possibility, only one respondent suggested that it had been a deciding factor in their choices. Considerations mentioned by respondents included: the need for caregivers; the inability to work (and associated loss of income and production); and the cost of adjusting one's home to accommodate a disabled person.

Q11. Might your answers have been different if you had been asked to choose which of two groups of patients to allocate health care funding to, as opposed to which of two patients to treat?

Most respondents said that considering groups would make no difference – the principles behind their decision-making would remain the same. Some suggested that it would be easier to make decisions when considering groups, as this would make the scenario more impersonal and therefore less 'heart-wrenching'. Others suggested that there would be a case for dividing resources amongst groups (with an even or uneven split), something that was not possible in the single treatment, two patient scenario.

Q12. "Society has a special responsibility towards those who have a short time left to live." What are your thoughts about this statement?

Most respondents agreed with this statement, though several were uncertain about the definitions of the terms 'society' and 'special'. A few respondents strongly disagreed with the statement, claiming that those with a short time left to live are no more important or worthy of treatment than anyone else. Several respondents claimed that caring for those at the end of life is not exclusively a health care system responsibility, arguing that society should focus on palliative care and objectives relating to ensuring sociability, comfort and dignity, rather than on life-extending health care technologies.

3.4 Discussion

This study piloted an approach to eliciting public preferences regarding life-extending end of life treatments. The purpose of the study was not to elicit preferences that can readily be used for decision-making, but to inform the design and methods of larger-scale, representative sample studies. The study was completed without any major problems, and offers support for the use of face-to-face interviews for this type of survey. The results provide some indication of public support for a policy that prioritises the treatment of patients with short life expectancy and whose disease onset has been sudden. However, all findings should be interpreted with caution, given the small sample size and exploratory nature of the study. The study highlighted a number of issues that warrant further investigation, some of which are examined below.

3.4.1 Unexpected rationales for choices

The comments made by respondents as they completed the survey indicate that a number of rationales exist for making priority-setting choices that had not been anticipated by the author. For example, it had not been anticipated that any respondents would prefer to treat patient A in S1. Yet three respondents did, arguing that it would be more valuable to give a life extension to someone who has more time to participate in society and who has a better opportunity to get their life in order. The fact that these respondents did not consider six months to be a worthwhile life extension for individuals with one year left to live indicates that they would also consider the three-month minimum standard applied in NICE's criteria to be insufficient.

It was hypothesised that if respondents switched from choosing to treat patient B in S1 to having no preference in S2, then the S1 choice will have been driven by time and/or age-related preferences. In fact, the two respondents who made this switch gave a different explanation, appearing to treat the additional time that patient A has as a 'sunk benefit'. Their argument was that if a benefit is yet to be enjoyed then it should be taken into account, but it has already passed then it should not. It had not been anticipated that respondents would apply a sunk benefit concept in their decision-making.

Finally, it had been anticipated that those respondents who used age information to decide which patient to treat would tend to choose to treat the patient who would be younger at their time of death. This would be consistent with a fair innings type argument (Williams, 1997) since the patient who would die younger in absence of treatment will have enjoyed a smaller amount of lifetime health, *ceteris paribus*. In

fact, some respondents sought to give priority to the patient who was younger *at the time of treatment*.

These examples show that people support a variety of principles of justice and rationing that may not be immediately obvious to researchers. It is therefore important to capture not only respondents' choices but also the reasons for those choices in order to understand fully the nature of their preferences.

3.4.2 Having no preference

Very few respondents indicated they had no preference between treating patient A and patient B. This (lack of) preference accounted for only 13% of all choices made. It may be the case that respondents' observed reluctance to express indifference was driven by the survey design. Beneath the written description and diagram for each scenario in the paper questionnaire (see Appendix 4), the prompt 'Would you prefer to treat patient A or patient B?' was presented. Although the interviewer made efforts to emphasise that a 'no preference' choice was acceptable, both at the beginning of the interview and whenever respondents displayed uncertainty, the fact that this option was not included in the written instructions may have resulted in a framing effect whereby it was deemed by respondents to be a unconventional response. It is recommended that in future studies any 'no preference' option should be included explicitly and given the same weight as competing options in any written and oral instructions. It is important to include a 'no preference' option, as this represents a defensible position for respondents who believe that end of life treatments should be given neither higher nor lower priority than any other treatments.

3.4.3 Interpreting '50% health'

Some respondents appeared to find the concept of '50% health' confusing. The reason that a percentage weight was used rather than a qualitative label was to minimise ambiguity about the severity of the imperfect health state in relation to perfect health. In practice, however, the way in which '50% health' was interpreted also differed from respondent to respondent. When describing their reasons for choosing to treat patient A in S4, some respondents appeared to be valuing one year in full health more highly than two years in 50% health, and it was not always clear whether this was based on a social preference or on a belief that the former is more desirable than the latter for the patients themselves. A possible solution would to emphasise the fact that 50% health should be interpreted as half as desirable as perfect health – for example, by adding the sentence: 'Patients have

told us that being in this health state for two years is equally desirable as being in full health for one year; we will therefore call this 50% health'.

3.5 Conclusions

This chapter has described an exploratory study that developed and piloted an approach for examining public support for giving higher priority to life-extending end of life treatments than to other types of treatments. The study was conducted successfully – respondents claimed to have found the survey interesting and the questions easy to understand. This suggests that a similar approach can be used for a larger study using a sample that is more representative of the general population.

Some aspects of the study design need refining, such as the explicit inclusion of an indifference option and the provision of guidance on how to interpret the concept of 50% health. Further, in order to inform the research question of this thesis, it is necessary to use a general public sample (rather than a convenience sample comprising university staff and students) and to collect sufficient data to be able to assess quantitatively the extent of support for an end of life premium. These limitations are addressed in empirical study 2 (Chapter 4).

4 VALUING HEALTH AT THE END OF LIFE: AN EMPIRICAL STUDY OF PUBLIC PREFERENCES (EMPIRICAL STUDY 2)[16]

4.1 Introduction

This chapter describes the second of the four empirical studies, conducted in mid-2011 following the successful completion of empirical study 1 (Chapter 3). The aim of this study is to examine whether a policy of giving higher priority to life-extending end of life treatments (as specified by NICE) than to other types of treatments is consistent with the preferences of members of the public.

Specific objectives are to test the following null hypotheses:

- People place no more weight on a unit of health gain for end of life patients than on that for non-end of life patients, *ceteris paribus*.
- 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.
- Any observed preferences regarding an end of life premium are unaffected by time preference.
- 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.
- Any observed preferences between quality of life-improving and life-extending treatments for end of life patients are unaffected by the ages of the patients.

4.2 Methods

4.2.1 Survey instrument

To test the hypotheses listed above, a survey was used, an earlier version of which was developed and piloted in empirical study 1 (Chapter 3). Hence, many aspects of the survey in this study are similar or identical to the empirical study 1 survey and are therefore not described in detail in this chapter.

[16] A previous version of this chapter has been published as: Shah, K.K., Tsuchiya, A., Wailoo, A.J., 2014. Valuing health at the end of life: an empirical study of public preferences. *European Journal of Health Economics*, 15, 389-399.

The survey included six scenarios (S1 to S6), preceded by a warm-up scenario (S0). All respondents considered all scenarios, in the same order. As with empirical study 1, respondents were presented with written descriptions and diagrams showing two hypothetical individuals (patient A and patient B) who have been diagnosed with illness. Both patients could benefit from treatment but the respondents were asked to assume that the health service had enough funds to treat one but not both of them.

Each scenario comprised two tasks. The first task required respondents to indicate which of three statements best described their view:

- I would prefer the health service to treat patient A
- I have no preference
- I would prefer the health service to treat Patient B

All three response options were given equal visual prominence.

The second task required respondents to consider a list of 18 statements, each describing a possible reason for their choice in the first task (reproduced in Appendix 6). They were asked to indicate, by ticking the relevant boxes, which of those statements were consistent with their own reasons. This task is hereafter referred to as the 'tick-box task'.

The scenarios are replicated in full in Appendix 6 and summarised in Table 4-1. In S1, S2 and S3, treatment would extend the life of either patient A or patient B by six months (with certainty); the patients differed in terms of age and the amount of time between diagnosis and expected death. These scenarios did not examine quality of life – respondents were advised that the patients' illnesses were asymptomatic and that treatment would not affect their quality of life. S1, S2 and S3 in this study correspond to S1, S3 and S2 in empirical study 1, respectively (the ordering of S2 and S3 in the sequence of scenarios was reversed in this study).

In S4, S5 and S6, the illnesses were described as having a negative effect on quality of life, with both patients experiencing their final year of life at 50% of full health. The concept of '50% health' was explained to respondents as follows: 'Patients have told us that being in this health state for two years is equally desirable as being in full health for one year'. In these scenarios, treatment would restore patient A to full health (with no effect on life expectancy) or extend the life of patient B by one year (with no effect on quality of life). S4 and S5 in this study build on S4 in empirical study 1, adding information on the specific ages of the patients, while S6 in this study corresponds to S5 in empirical study 1.

Table 4-1. Summary of scenarios used in empirical study 2

	S1	S2	S3	S4	S5	S6
Without treatment						
Patient A life expectancy	10 years	10 years	1 year	1 year	1 year	10 years
Patient A quality of life	100%	100%	100%	50%	50%	50%
Patient B life expectancy	1 year	1 year	1 year	1 year	1 year	1 year
Patient B quality of life	100%	100%	100%	50%	50%	50%
Gains from treatment						
Patient A life expectancy	+6 months	+6 months	+6 months	No change	No change	No change
Patient A quality of life	No change	No change	No change	+50%	+50%	+50% [a]
Patient B life expectancy	+6 months	+6 months	+6 months	+1 year	+1 year	+1 year
Patient B quality of life	No change	No change	No change	No change	No change	No change
Undiscounted QALY gain from treatment [b]						
Patient A	0.5 QALY	0.5 QALY	0.5 QALY	0.5 QALY	0.5 QALY	0.5 QALY
Patient B	0.5 QALY	0.5 QALY	0.5 QALY	0.5 QALY	0.5 QALY	0.5 QALY
Age of patients	A & B are same age (adults)	B is 9 years older than A (adults)	A & B are same age (adults)	A & B are same age (30 years old)	A & B are same age (70 years old)	B is 9 years older than A (adults)
Timing of scenario (when does the treatment decision occur)	At time of A & B's diagnosis	At time of A & B's diagnosis	9 years after A's diagnosis	At time of A & B's diagnosis	At time of A & B's diagnosis	At time of A & B's diagnosis

[a] Refers to the quality of life improvement in the patient's final year of life
[b] Respondents did not see this information (the term 'QALY' was not used at any point in the survey)

Under the conditions that two years in 50% health is equivalent to one year in full health, and that a health gain today is equivalent to an equal-sized health gain in the future, both patients will gain exactly the same amount of health from treatment in all six scenarios – half of a QALY. Unlike in empirical study 1, a specific explanation of the meaning of 50% health was provided to respondents (see above).

Explanations of scenarios S1, S2 and S3 are provided in the description of empirical study 1 (see 3.2.1) and are not repeated here, though the reader is reminded that S2 in empirical study 1 corresponds to S3 in the current study, and *vice versa*. As mentioned above, considerations of quality of life are introduced in scenarios S4, S5 and S6. S4 and S5 involve choosing between treatments that extend life and treatments that improve quality of life. The two scenarios are identical except for the ages of the patients. In S4, both patients are younger adults (30 years old). In S5, they are older adults (70 years old). Comparing the results of S4 and S5 therefore provides an indication of whether the preference for a particular type of treatment (life-extending or quality of life-improving) for end of life patients is dependent on the life stage of the patients. Scenario S6 combines elements of S2 and S4/S5 in that it involves choosing between treating a non-end of life patient

(patient A) and an end of life patient (patient B) and between a quality of life-improving treatment (to patient A) and a life-extending treatment (to patient B).

It is conjectured that a supporter of NICE's end of life policy would choose to treat patient B in all scenarios, except perhaps S3 where it is less clear whether the supplementary advice applies.

Warm-up scenario S0 involved choosing between giving a two-year life extension to a patient with 10 years of life expectancy in full health and a six-month life extension to a patient with one year of life expectancy in full health. S0 was included as a practice task to familiarise the respondent with the priority-setting exercise. The responses for S0 have not been analysed and are not reported here.

4.2.2 Administration of survey

The survey was administered using face-to-face interviews, undertaken by a team of six interviewers employed by a market research agency, Accent. The interviewers completed training on the specifics of the methodology and procedures for this study. All interviews were carried out in a one-to-one setting in the homes of respondents.

Background information (age group, gender, social grade) was collected at the beginning of the interview. Respondents then considered the scenarios one at a time, progressing to the next scenario once they had been given time to consider their views and had provided answers to the questions. The interviewers permitted respondents to amend their responses to earlier questions if they changed their mind during the course of the interview.

Information about the scenarios was presented in three ways: (1) the full scenario description was read aloud to the respondent by the interviewer, following a script; (2) key pieces of information were presented schematically using a diagram; and (3) key pieces of information were presented in a summary table beneath the diagram. The interview materials are reproduced in Appendix 6. The diagrams and summary tables were included in a paper booklet handed to each respondent. These booklets were also used to record respondents' answers to the questions.

After concluding the interview, the interviewer was asked to answer three 'diagnostic' multiple choice questions. These were concerned with assessing: (1) how well the respondent had understood and carried out the tasks; (2) how much effort and concentration the respondent had put into the tasks; and (3) the extent to which there were disruptions and interruptions in the interview environment.

All data were collated and entered into an Excel database by the agency.

4.2.3 Sample

The survey was administered on a sample of adult members of the general public, split evenly between two areas of southeast England (London and Kent). The target sample size of 50 respondents was determined on the basis of available resources. A 'minimum quota' approach was used to recruit a sample that was broadly representative of the general population in terms of age, gender and social grade. The sample was recruited using a 'door knock' approach, with the interviewer approaching a household member of every fourth home in a randomly allocated postal area and scheduling interview appointments for those individuals that agreed to participate. A small cash payment was offered as an incentive for participation.

4.2.4 Ethical approval

The survey design 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.

4.2.5 Piloting

As mentioned above, the survey used in this study was developed and piloted in empirical study 1 (Chapter 3). The pilot was completed successfully, indicating that a similar approach could be used for a larger study using a general public sample.

The findings from empirical study 1 informed the design of the current study in a number of ways, in particular: the inclusion of a warm-up scenario; the specification that both patients are adults; the inclusion of two scenarios (S4 and S5) exploring the extent to which preferences regarding end of life treatments are driven by considerations of the 'life stage' of patients; the reversal of the order in which S2 and S3 appeared in the sequence of scenarios; equal visual prominence for all three response options in respondents' answer booklets; increased clarity in the description of what is meant by '50% health'; and changes to the ways in which information about the scenarios was presented to respondents. The list of statements included in the tick-box task for each scenario was developed in accordance with the qualitative data obtained in the pilot.

4.2.6 Methods of analysis

Descriptive statistics were reported in order to summarise the sample and responses to the scenario questions. Comparisons 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 B in one scenario and choosing to treat patient B in the other scenario. 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. Probability values (p-values) of the test statistics were used to assess the strength of the evidence against the null hypotheses (Fisher, 1956). P-values below 0.05 were considered 'strong' evidence against the null hypothesis, while p-values between 0.05 and 0.1 were considered 'weak' evidence.

For each scenario, a list of inconsistent sets of responses (either a tick-box response that contradicts a given choice task response; or a tick-box response that contradicts another tick-box response) was generated. The number of respondents whose data contained at least one inconsistent set of responses was counted.

4.3 Results

Interviews were completed by 50 respondents in July 2011. The background characteristics of the sample are presented in Table 4-2. The sample includes a larger proportion of older individuals, and a smaller proportion of middle-aged individuals, than in the general population (Office for National Statistics, 2011). The sample also includes a relatively large proportion of individuals in the lowest social grades (National Readership Survey, 2012-3).

Table 4-2. Sample background characteristics

		n	%	Population
Total		50	100%	
Age (years)	18-34	14	28%	29%
	35-64	18	36%	50%
	65+	18	36%	21%
Gender	Female	24	48%	51%
	Male	26	52%	49%
Social grade [a]	A	1	2%	4%
	B	7	14%	22%
	C1	17	34%	29%
	C2	6	12%	21%
	DE	19	38%	23%

[a] Refers to the occupation/qualifications/responsibilities of the chief wage earner of the respondent's household; see National Readership Survey (2012-13).

All 50 respondents completed the survey in full. According to the interviewers, the majority of interviews were carried out in distraction-free environments with respondents who concentrated on and showed a good understanding of the survey tasks.

4.3.1 Response data

Table 4-3 reports aggregate response data for each of the six scenarios.

Table 4-3. Aggregate response data for all scenarios

	S1	S2	S3	S4	S5	S6
Prefer to treat patient A	13 (26%)	16 (32%)	16 (32%)	29 (58%)	28 (56%)	31 (62%)
No preference	7 (14%)	12 (24%)	13 (26%)	10 (20%)	11 (22%)	7 (14%)
Prefer to treat patient B	30 (60%)	22 (44%)	21 (42%)	11 (22%)	11 (22%)	12 (13%)
Total	50 (100%)	50 (100%)	50 (100%)	50 (100%)	50 (100%)	50 (100%)

In S1, S2 and S3, preferring to treat patient B (the patient whose life expectancy is shorter or who has known about their illness for less time) was the most popular choice, although it is only in S1 that the majority of respondents (60%) made this choice. In all three scenarios, there were more respondents who preferred to treat patient A than there were respondents who indicated that they had no preference between the two patients. The proportion of respondents choosing to treat the end of life patient in S1 is statistically significantly greater than 50% at the 10% level of confidence but not at the 5% level (binomial test using normal approximation; p=0.08). Hence, the hypothesis that people place no more weight on a unit of health gain for end of life patients than on that for non-end of life patients (hypothesis 1) is *rejected*.

In S4 and S5, the majority of respondents (58% and 56%, respectively) preferred to treat patient A, for whom treatment would deliver a quality of life improvement worth half a QALY. The remainder of respondents were roughly evenly split between preferring to treat patient B – for whom treatment would deliver a life extension worth half a QALY – and having no preference. In S6, the majority of respondents (62%) preferred to treat patient A, which involved giving a quality of life improvement worth half a QALY to the non-end of life patient.

Table 4-4 to Table 4-7 provide cross-tabulations of the response data from selected combinations of scenarios that can be used to test the hypotheses set out in 4.1. In these tables, the values in each cell refers to the number and proportion of respondents expressing that set of preferences.

The most common pair of choices in S1 and S2 was to prefer to treat patient B in both scenarios (made by 19 respondents – 38%) (Table 4-4). The most common reasons given in the tick-box task for preferring to treat patient B were that this choice benefits the patient 'who is closest to death' and who has 'less time to

prepare for death'. Four respondents (8%) switched from preferring to treat patient B in S1 to having no preference in S2. Three of these respondents indicated in the tick-box task for S1 that their choice 'benefits the patient who will die at a younger age'. A further seven respondents (14%) preferred to treat patient B in S1 and to treat patient A in S2. There does not appear to be a consensus set of reasons given for this pair of choices. Three of these respondents did not give any reasons linked to the ages of patients; of the four that *did* give reasons linked to age, two gave reasons that were factually incorrect (for example, stating that treating patient A in S2 involved benefiting the patient 'who will die at an older age').

The second most common pair of choices was to prefer to treat patient A in both scenarios (made by eight respondents – 16%). Almost all of the respondents who chose to treat patient A indicated in the tick-box tasks that they did so because they wished to benefit the patient 'who has longer left to live'. Overall, the association between patient age and the propensity to favour the treatment of the end of life patient is not statistically significant (chi-squared test; p=0.16). 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) *cannot be rejected*.

Table 4-4. Cross-tabulation – S1 versus S2

	S2			
S1	Prefer A	No preference	Prefer B	Total
Prefer A	8 (16%)	3 (6%)	2 (4%)	13 (26%)
No preference	1 (2%)	5 (10%)	1 (2%)	7 (14%)
Prefer B	7 (14%)	4 (8%)	19 (38%)	30 (60%)
Total	16 (32%)	12 (24%)	22 (44%)	50 (100%)

In S1, patient A and patient B are the same age today, so without treatment patient A will be nine years older when they die than patient B will be when they die. In S2, patient B is nine years older than patient A, so without treatment both patients will be the same age when they die.

The most common pair of choices in S2 and S3 was to prefer to treat patient B in both scenarios (made by 10 respondents – 20%) (Table 4-5). The majority of the respondents who made this pair of choices ticked the box that read 'My choice benefits the patient with less time to prepare for death' for both scenarios. Eight of the 10 respondents ticked the box that read 'My choice benefits the patient who is closest to death' for S2, whilst three gave the equivalent reason for S3, when the statement was factually incorrect.

Five respondents (10%) preferred to treat patient B in S2 and had no preference in S3. Of these respondents, only one ticked the box for S2 that read 'My choice delivers the benefit today rather than far away in the future'. More popular reasons given for preferring to treat patient B in S2 were that patient B: 'is older today',

'has less time to prepare for death', and/or 'can make the most of their remaining time'.

Seven respondents (14%) preferred to treat patient B in S2 and to treat patient A in S3. There does not appear to be a consensus set of reasons given for this pair of choices. The most commonly given reason was that preferring to treat patient B in S2 'delivers the benefit today rather than far away in the future' (three of the seven respondents ticked this box). A further seven respondents (14%) preferred to treat patient A in S2 and to treat patient B in S3. The only reason that was given consistently for this pair of choices was that patient A has 'longer left to live' in S2 (six of the seven respondents ticked this box).

Overall, there is no statistically significant evidence of an association between the timing of the scenario and the propensity to favour the treatment of patient B (chi-squared test; p=1.00). Hence, the hypothesis that preferences regarding an end of life premium are unaffected by time preference (hypothesis 3) *cannot be rejected*.

Table 4-5. Cross-tabulation – S2 versus S3

	S3			
S2	Prefer A	No preference	Prefer B	Total
Prefer A	6 (12%)	3 (6%)	7 (14%)	16 (32%)
No preference	3 (6%)	5 (10%)	4 (8%)	12 (24%)
Prefer B	7 (14%)	5 (10%)	10 (20%)	22 (44%)
Total	16 (32%)	13 (26%)	21 (42%)	50 (100%)

In S2, the treatment decision occurs at the time of diagnosis for both patient A and patient B. In S3, the treatment decision occurs at the time of diagnosis for patient B and nine years after the time of diagnosis for patient A.

The majority of respondents made the same choice in both S4 and S5 – 22 respondents (44%) preferred to treat patient A in both scenarios; 8 respondents (16%) expressed no preference in both scenarios; and six respondents (12%) preferred to treat patient B in both scenarios (Table 4-6). Five respondents (10%) preferred to treat patient B in S4 and to treat patient A in S5. Four respondents (8%) made the reverse pair of choices, preferring to treat patient A in S4 and to treat patient B in S5. Some, but less than half, of the respondents who made each pair of choices gave reasons that were consistent with those choices – i.e. by ticking the relevant box that read 'I think it is better to [improve health/extend life] than to [extend life/improve health] in this situation'. Another commonly given reason (particularly for respondents who preferred to treat patient A in S4 and to treat patient B in S5) was that their choice 'benefits the patient who can make the most out of their remaining time'.

Considering all of the 14 respondents who did *not* make the same choice in both S4 and S5, there was no consensus as to whether it is preferable to give a quality of

life improvement to a younger adult and a life extension to an older adult, or *vice versa*. In fact, the data suggest a fairly even split between these two views.

Overall, the proportion of respondents choosing to provide the life-extending treatment is statistically significantly less than 50% (binomial test using normal approximation; $p<0.01$). 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*. However, there is no statistically significant evidence of an association between the life stage of the patient and the propensity to choose either of these types of treatment (chi-square test of association; $p=0.97$). Hence, the hypothesis that preferences between quality of life-improving and life-extending treatments for end of life patients are unaffected by the ages of the patients (hypothesis 5) *cannot be rejected*.

Table 4-6. Cross-tabulation – S4 versus S5

	S5			
S4	Prefer A	No preference	Prefer B	Total
Prefer A	22 (44%)	3 (6%)	4 (8%)	29 (59%)
No preference	1 (2%)	8 (16%)	1 (2%)	10 (20%)
Prefer B	5 (10%)	0 (0%)	6 (12%)	11 (22%)
Total	28 (56%)	11 (22%)	11 (22%)	50 (100%)

In S4, both patients are 30 years old. In S5, both patients are 70 years old.

The most common pair of choices in S2 and S6 was to prefer to treat patient B in S2 and to treat patient A in S6 (made by 15 respondents – 30%) (Table 4-7). The most commonly given reasons for this pair of choices were that treating patient B in S2 'benefits the patient with less time to prepare for death'; and that 'it is better to improve health than extend life' in the situation depicted in S6 (both boxes were ticked by seven of the 15 respondents). A small number of respondents ticked boxes relating to age and/or the timing of the benefits to explain their choices. Overall, there is statistically significant evidence of an association between the availability of quality of life-improving treatment and the propensity to give priority to the life-extending end of life treatment at the 10% level of confidence, but not at the 5% level (chi-squared test; $p=0.06$).

Table 4-7. Cross-tabulation – S2 versus S6

	S6			
S2	Prefer A	No preference	Prefer B	Total
Prefer A	10 (20%)	2 (4%)	4 (8%)	16 (32%)
No preference	6 (12%)	5 (10%)	1 (2%)	12 (24%)
Prefer B	15 (30%)	0 (0%)	7 (14%)	22 (44%)
Total	31 (62%)	7 (14%)	12 (24%)	50 (100%)

In S2, the choice is between giving a life extension worth ½ QALY to the non-end of life patient (patient A) and a life extension worth ½ QALY to the end of life patient. In S6, the choice is between giving a quality of life. improvement worth ½ QALY to the non-end of life patient (patient A) and a life extension worth ½ QALY to the end of life patient (patient B).

Across the six scenarios, there were 70 instances of respondents giving reasons in the tick-box task that were inconsistent with their choice or with other reasons they gave for making the choice in the same scenario. More than half of these observations came from six respondents who each failed the 'consistency checks' on four or more occasions. The majority of respondents did not fail any of these checks. The 'None of the above' box was ticked on six occasions across all scenarios, five of which were for the tick-box task for S3. In all cases, respondents who ticked the 'None of the above' box did not tick boxes describing any other reasons.

Considering the various combinations of choices made across all six scenarios, 39 different sets of choices were made by the 50 respondents. The most popular set of choices (BBBAAA) was made by four respondents (8%); most sets were made by only one respondent.[17] Three respondents expressed no preference throughout the six scenarios (IIIIII). No respondents made the set(s) of choices implied by the NICE end of life policy of giving greater priority to life-extending end of life treatments than to both non-end of life treatments and quality of life-improving end of life treatments (BBBBBB or BBIBBB).

4.4 Discussion

This study elicited the preferences of a sample of 50 members of the general public in England over health care priority-setting scenarios. Whilst the results should be interpreted with a degree of caution given the limited size and geographic coverage of the sample, they provide a number of insights on whether there is public support for a policy that places greater weight on life-extending end of life treatments (as specified by NICE) than on other types of treatments.

The results suggest that: (1) there is weak evidence that people place greater weight on the treatment of end of life patients than on the treatment of non-end of life patients; (2) there is no evidence that concern about age is a motivating factor for giving higher priority to the treatment of end of life patients; (3) there is no evidence that time preference is a motivating factor for giving higher priority to the treatment of end of life patients; (4) there is strong evidence that people do not place greater weight on life-extending than on quality of life-improving treatments for end of life patients; and (5) there is no evidence that concern about the life stage of end of life patients is a motivating factor for preferring either life-extending or quality of life-improving treatments for those patients.

[17] Note that in these sets of choices, A (B) indicates that the respondent preferred to treat patient A (B), and I indicates that the respondent had no preference

The fact that the most popular choice was to treat patient B in S1 and S2 indicates support for prioritising the treatment of patients with short life expectancy, but in both cases there is no statistically significant evidence at the 5% level that the majority of respondents held this view. Moreover, a non-trivial minority of respondents indicated the opposite – that is, they preferred to give higher priority to the treatment of patients with *longer* life expectancy. Data from the tick-box tasks suggest that such preferences may be driven by a belief that patients with longer life expectancy are better placed to make the most out of a short life extension.

The most popular choice in S3 was also to treat patient B. This preference may be driven by concern about how much time the patient has had to 'prepare for death'. Since patient B's disease progression has been more sudden, they have had less time to prepare for death. The reasons given for choosing to treat patient B in the tick-box task for S3 support this explanation.

The response data for S4 and S5 provide evidence of an overall preference for giving priority to quality of life-improving rather than life-extending treatments for patients with short life expectancy. Furthermore, the data for S6 suggest that some respondents prefer to give priority to quality of life-improving over life-extending treatments even when the quality of life improvement accrues to a non-end of life patient and the life extension (of equal size, in QALY terms) accrues to an end of life patient. One interpretation of these results is that the preference for giving priority to quality of life-improving treatments is so strong that it outweighs any preference for giving priority to end of life treatments. Another interpretation is that the respondents have misunderstood (or rejected) the concept of the QALY, believing that a half-QALY quality of life improvement is in fact more desirable than a half-QALY life extension for the patients themselves. This issue is likely to exist in any study where quality of life needs to be quantified in this manner.

Comparing the response data for S1 and S2 allowed an assessment of whether respondents' preferences for treating the end of life or the non-end of life patient depended on the ages at which the patients would die without treatment. Similarly, comparing the response data for S4 and S5 allowed an assessment of whether respondents' preferences for either life-extending or quality of life-improving treatments for end of life patients depended on the life stage of the patients (i.e. whether they were younger adults or older adults). In both cases, the particular combinations of ages that were used in the scenarios do not appear to have influenced the choices of most of the respondents. However, concerns about age might have been more evident had the difference in ages been greater in S1 and

S2, or if different life stages had been presented in S4 and S5 (for example, if the patients were described as teenagers aged 15 years in S4 and as elderly individuals aged 90 years in S5).

In all six scenarios, the least popular choice was for respondents to express no preference between treating patient A and patient B. This is consistent with findings from empirical study 1 (see 3.4.2) and adds to the evidence that people do not support a strict health-maximisation objective when making priority-setting choices (Dolan *et al.*, 2005), choosing instead to prioritise based on characteristics of the patients, diseases or treatments under consideration. It may be the case, however, that for some respondents health-maximisation is the *primary* objective, but when there is nothing to choose between the two patients in terms of (undiscounted) health gain, they refer to other factors (such as life expectancy without treatment) as a 'tie breaker'. Following concerns about framing effects in empirical study 1, steps were taken to make the 'no preference' option explicit and prominent in this study so as to make it clear that this was an acceptable response and that respondents were not obliged to choose to treat patient A or patient B. The tick-box task for each scenario also included several statements referring to reasons for having no preference. However, most respondents still preferred to treat one patient or the other, and in general provided reasons in the tick-box tasks that were consistent with their choices.

There cannot be described to be a 'consensus' combination of preferences – the most popular set of choices (BBBAAA) was made by only four respondents (8%). This set of choices indicates a preference for giving priority to those at the end of life (preferring to treat patient B in S1 and S2) and whose disease progression has been sudden (preferring to treat patient B in S3); a preference for quality of life-improving rather than life-extending treatments (preferring to treat patient A in S4 and S5); and a preference for quality of life-improving treatments that outweighs the preference for giving priority to those at the end of life (preferring to treat patient A in S6).

It is difficult to describe a single approach to priority-setting that reflects the heterogeneous preferences elicited in this study. It is of note, however, that no respondents made the set of choices implied by the NICE end of life policy of giving greater priority to life-extending end of life treatments than to both non-end of life treatments and quality of life-improving end of life treatments (BBBBB or BBIBBB).

The study results suggest that the NICE's policy may be insufficient in two ways. First, whilst it is concerned with patients' remaining life years, the supplementary advice does not distinguish between sudden and non-sudden disease progression.

Findings from empirical study 1 (see 3.3.2), coupled with an examination of the reasons given by respondents in the tick-box tasks in this study, suggest that for many people the preference for prioritising the treatment of end of life patients is driven by concern about how much time the patients will have had to prepare for death. This may explain why over 40% of respondents preferred to treat patient B in S3, despite the fact that patient B is no closer to their end of life than patient A in that scenario. Nevertheless, it should be noted that no respondents made the BBBBBB set of choices (which implies support for an adjusted NICE policy that also gives priority to patients whose disease progression has been most sudden). It should also be noted that of the six occasions when respondents ticked the box that read 'None of the above' when providing reasons for their choices, five referred to their choice in S3. This suggests that there are rationales for choosing either patient A or patient B in S3 beyond those that had been conjectured, and that more work is needed to better understand people's preferences regarding prioritisation according to the speed of disease progression.

Second, the NICE policy involves giving greater weight to life-extending but not to quality of life-improving treatments for those at the end of life. This is inconsistent with the finding in this study that many respondents favoured the prioritisation of the quality of life-improving treatment over the life-extending treatment in S4, S5 and S6.

Whilst the results of this study provide an indication of whether there is public support for the policy of giving higher priority to life-extending end of life treatments than to other types of treatments, they do not give any indication of the *strength* of preferences for any individual respondent, nor do they indicate whether the cut-offs in the current NICE criteria (for example, defining short life expectancy as 'normally less than 24 months') are commensurate with public preferences.

Nevertheless, there are a number of ways in which the findings of this study can help inform the design of future studies of public preferences regarding the value of end of life treatments. A key challenge for stated preference studies is defining attributes and levels that are policy relevant and salient to respondents (Lancsar and Louviere, 2008). This study has provided evidence to suggest that the amount of time the patient has had to prepare for death and the nature of the health gain offered by the treatment (quality of life-improving or life-extending) are key drivers of preferences. It is therefore recommended that future studies should seek to better understand the strength of preferences regarding these attributes. On the other hand, this study provided little evidence to suggest that respondents' preferences regarding end of life treatments are influenced by the age of the

patients. However, it should be borne in mind that preferences were sought only regarding younger adults and older adults – scenarios examining children, for example, may well have generated different results. It is necessary to include a greater variety of levels for attributes such as life expectancy in order to understand whether those attributes exhibit non-linear or threshold effects on respondents' preferences. For instance, people may prefer to give priority to patients with shorter life expectancy up to a point, but when the life expectancy becomes extremely short (for example, less than three months) then this preference may no longer hold.

In this study, all of the scenarios were designed such that both patients received the same amount of undiscounted health gain – half a QALY in all cases. In reality, the NICE end of life policy has led to a situation whereby some end of life treatments offering very small improvements in health have been recommended for use in the NHS (Longson and Littejohns, 2009), whilst non-end of life treatments offering much larger benefits have not been recommended. It would therefore be useful to understand the extent to which people are willing to sacrifice overall health benefit in order to give priority to the treatment of end of life patients. This could be examined by varying the levels of the 'health gain from treatment' attribute.

Public preference studies can help develop an understanding of what people consider to be appropriate and acceptable for society. In order to obtain a comprehensive picture, however, it is important to capture not only respondents' choices but also the reasons for their choices. The tick-box tasks were valuable in this respect, but were limited in that many respondents simply ticked boxes that referred to 'factually correct' statements but did not necessarily offer any insight into the nature of their preferences. Moreover, a large number of respondents gave reasons that were inconsistent with each other or with their choices. Future studies should consider alternative ways of eliciting information to aid the interpretation of observed preferences.

4.5 Conclusions

This chapter has described a small-scale study that examined whether the policy of giving higher priority to life-extending end of life treatments (as specified by NICE) than to other types of treatments is consistent with the preferences of members of the general public. The results provide some weak evidence of public support for giving priority to patients with relatively short life expectancy, but it should be noted that a sizeable minority of respondents expressed the opposite preference. The current NICE policy does not cover quality of life-improving end of life

treatments, and is not concerned with whether the treatments under appraisal are indicated for patients whose disease progression has been sudden. Yet the results of this study suggest that people's preferences regarding the value of end of life treatments may be influenced by these factors.

Given the small sample, limited range of scenarios, and fragility of some of the results, the findings of this chapter should not be taken as definitive. A larger scale study, designed to examine more robustly people's preferences regarding end of life treatments, is recommended. Empirical study 3 (Chapter 5) addresses some of the limitations described above. By varying the gains from treatment across alternatives and choice tasks, empirical study 3 provides information about the trade-off between health-maximisation and giving priority to end of life treatments. It also investigates the extent to which the cut-offs in the NICE criteria are consistent with public preferences.

5 VALUING HEALTH AT THE END OF LIFE: A STATED PREFERENCE DISCRETE CHOICE EXPERIMENT (EMPIRICAL STUDY 3)[18]

5.1 Introduction

This chapter describes the third of the four empirical studies, conducted in 2012. At that time, evidence of public preferences regarding NICE's policy remained scarce. Empirical study 2 (Chapter 4) had provided weak and tentative evidence of support for an end of life premium, and had concluded with a recommendation to undertake a more robust and larger scale examination of end of life-related preferences. Empirical study 3 advances the previous studies by exploring whether and how people would sacrifice overall health gains in order to give priority to end of life patients, and by testing alternative operationalisations of NICE's policy.

As in empirical study 2, the aim is to examine whether the policy of giving higher priority to life-extending end of life treatments (as defined by NICE) than to other types of treatments is consistent with the preferences of members of the general public. A large-scale web-based DCE is used to address this research question. The study is one of the first to have applied the DCE method to examine preferences in the end of life context. The predicted probability approach used by Green and Gerard (2009) is followed in order to present results on a probability scale with ratio level properties, thereby allowing an assessment of the strength of preferences between competing profiles or scenarios.

A secondary objective is to examine further the question of whether people's preferences regarding the treatment of end of life patients are affected by how long those patients have known about their prognosis.

5.2 Methods

5.2.1 Survey instrument

5.2.1.1 Framework

There are many stated preference techniques that can be used to elicit public preferences regarding health care priority-setting (Ryan *et al.*, 2001). Health economists typically prefer choice-based methods that reflect the view that the value of something is measured by how much one is willing to trade or sacrifice to

[18] A previous version of this chapter has been published as: Shah, K.K., Tsuchiya, A. and Wailoo, A.J., 2015. Valuing health at the end of life: a stated preference discrete choice experiment. *Social Science and Medicine* 124, 48-56.

obtain it. The literature review identified eight types of choice-based methods that have been used in studies reporting evidence on preferences regarding end of life treatments (see 2.3.4), including simple choice exercises as used in empirical studies 1 and 2 in this thesis (Chapters 3 and 4, respectively).

A specific variant of choice exercise, the DCE, produces quantitative trade-offs between different factors based on hypothetical choices (Louviere *et al.*, 2000). DCEs are typically implemented in surveys comprising several 'choice sets', each containing competing alternative 'profiles' described using 'attributes' and a range of attribute 'levels'. Respondents are asked to choose between these alternative profiles, and the resulting choices are analysed to estimate the relative contribution of each of the attribute levels to overall utility (Lancsar and Louviere, 2008).

DCE data are modelled within a random utility framework, which assumes the utility (U_{nj}) that respondent *n* obtains from choosing alternative *j* can be separated into an explainable component (V_{nj}) and an unexplainable component (ε_{nj}):

$$U_{nj} = V_{nj} + \varepsilon_{nj}$$

The researcher does not observe ε_{nj} and treats it as random. V_{nj} is the indirect utility function in which the attributes of the alternatives are arguments. The probability that the respondent chooses alternative *i* over alternative *j* is given by:

$$P_{ni} = \Pr\left(V_{ni} + \varepsilon_{ni} > V_{nj} + \varepsilon_{nj}\right) \quad \forall j \neq i$$
$$= \Pr\left(V_{ni} - V_{nj} > \varepsilon_{nj} - \varepsilon_{ni}\right) \quad \forall j \neq i$$

Assuming that the random terms are independently and identically distributed, the conditional logit model can be used to derive probability outcomes across a choice set (Louviere *et al.*, 2000). The predicted probability of alternative *i* being chosen from the complete set of alternatives (*j*=1,…,J) is given by:

$$P_{ni} = \frac{e^{V_{ni}}}{\sum_{j=1}^{\mathrm{J}} e^{V_{nj}}} \quad j = 1, \ldots, \mathrm{J}$$

The number of studies using DCEs in health economics has grown rapidly in recent years (de Bekker-Grob *et al.*, 2012; Clark *et al.*, 2014), though until the early 2000s most applications had been concerned with eliciting individual personal preferences from respondents who had been asked to consider the choice context as it applied to themselves (Green and Gerard, 2009). An increasing trend, however, is to use DCEs to examine social preferences whereby respondents are asked to adopt a social decision-maker perspective and consider choices involving other people in society (Whitty *et al.*, 2014). This is the context adopted in this study.

5.2.1.2 Attributes and levels

The selection of attributes and levels (Table 5-1) was based on NICE's criteria (see 1.8.2) and informed by the findings of empirical studies 1 and 2. 'Life expectancy without treatment' and 'life expectancy gain from treatment' were included as attributes as these form the basis for the critera in NICE's end of life policy. The levels for these attributes were selected so as to examine whether there is a case for amending the cut-offs implied by the existing criteria. For life expectancy without treatment, a level representing the current cut-off of 24 months was included, as well as two levels smaller and two levels larger than this cut-off (three months, 12 months; 36 months, 60 months). An even larger level of 120 months (or 10 years) was considered but omitted due to concerns about how the durations would be displayed visually using computer-based diagrams (see Figure 5-1). Similarly, the current 'life expectancy gain from treatment' cut-off of three months was included, as well as two smaller and two larger levels (one month, two months; six months, 12 months). In addition, 0 months was included in order to examine preferences for end of life treatments that offer no life extension.

Table 5-1. Attributes and levels used in the study

Attribute	Unit	Levels
Life expectancy without treatment	months	3, 12, 24, 36, 60
Quality of life without treatment	%	50, 100
Life expectancy gain from treatment	months	0, 1, 2, 3, 6, 12
Quality of life gain from treatment	%	0, 25, 50

The inclusion of quality of life attributes was driven by the finding in empirical studies 1 and 2 that many respondents appeared to favour the prioritisation of quality of life-improving treatments over life-extending treatments for end of life patients. The term 'quality of life' was not used in the survey itself; following Rowen *et al.* (2016a), this attribute was described using a health scale ranging from 'dead' (0%) to 'full health' (100%).

Whilst other studies have presented quality of life using a wide range of levels – see Baker *et al.* (2010a), for example – the piloting work in empirical study 1 indicated that this may be challenging for respondents to interpret (see 3.4.3). Hence, only two levels were selected for the 'quality of life without treatment' attribute: 50% and 100%. The concept of '50% health' was explained in the instructions as follows (consistent with the instruction provided to respondents in empirical study 2): '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.' The three levels for the 'quality of life gain from treatment' attribute

were designed to represent treatments that: (1) offer no health improvement (0% gain); (2) restore the patient to full health (50% gain); and (3) offer some improvement but do not restore the patient to full health (25% gain).

Other potential attributes, such as the patient's age or past health, were considered but eventually omitted from the final study design in order to restrict the complexity of the choice tasks. Whilst the literature is inconclusive with regard to the number of attributes that should be included in DCEs, some researchers have suggested that when tasks become too complex respondents may not make trade-offs but instead adopt other decision heuristics or lexicographic decision rules (Witt *et al.*, 2009). This study therefore followed a parsimonious approach, focusing on the attributes that are most salient to the policy context for NICE.

On the other hand, the results of empirical studies 1 and 2 suggested that people's preferences regarding end of life treatments may be guided by how long the patients have known about their illness or prognosis. A patient who has only just found out about their illness may be prioritised differently from one who has known about their illness for some time, even if both patients' life expectancies are similar (Table 4-5). A number of approaches for examining preferences regarding this issue were considered. Due to the complexities involved in incorporating a 'time with knowledge of illness' attribute into the experimental design, a pragmatic decision was made to restrict the attributes in the DCE tasks to those listed in Table 5-1, and to add two further 'extension' pairwise choice tasks to the survey which focused specifically on the impact of this additional attribute. These extension tasks do not form part of the experimental design for the DCE but were designed so as to enable direct comparisons with the corresponding 'standard' tasks. See 5.2.1.4 for details.

5.2.1.3 Experimental design

A full factorial design using the attributes and levels listed in Table 5-1 would have resulted in 5*2*6*3 = 180 possible profiles, but some combinations would result in implausible scenarios. The sum of quality of life without treatment and quality of life gain from treatment cannot exceed 100% as it is not possible to have a health state that is better than full health. A constraint that the sum of life expectancy gain from treatment and quality of life gain from treatment must be greater than zero was also imposed, or else the treatment would offer no improvement. Imposing these constraints suppressed 70 of the 180 possible profiles, leaving 110 profiles and 5,995 possible pairwise choices sets to select from.

Using the Stata software package (StataCorp, 2013), 80 pairwise choice sets were constructed from these 110 profiles using a D-optimality algorithm (Carlsson and Martinsson, 2003) with the attribute coefficients set to zero. The design allowed for the estimation of both main effects and selected interaction effects (see 5.2.6). All of the choice sets were checked for plausibility, and no manual alteration of the design was required.

There is little guidance in the literature on the optimal number of DCE tasks to ask each respondent to complete in a single survey. The social preference DCE studies summarised by Green and Gerard (2009) used between one and 18 choice sets per respondent, whilst in a review of 79 studies, Marshall *et al.* (2010) report that the majority of studies used between seven and 15 choice sets. The 80 choices sets in this study were organised into eight blocks of 10 choices. Each of the 80 choice sets was also assigned to one of 13 different 'choice types' according to the nature of the choice being depicted (Table 5-4). For example, in 10 of the 80 choice sets, the patient with shorter life expectancy without treatment gains more quality of life from treatment than the patient with longer life expectancy (choice type 3). Similarly, in 11 of the 80 choice sets, both patients have the same amount of life expectancy and quality of life without treatment, but one patient gains more life expectancy and more quality of life from treatment than the other (choice type 1). Assuming that that larger health gains should always be preferred to smaller health gains, *ceteris paribus* (an assumption that is inherent to a QALY-maximisation approach to resource allocation), choosing the patient who gains more life expectancy and quality of life from treatment can be regarded as the dominant option and should always be preferred. These choice sets therefore provide an opportunity to test whether respondents' preferences conform to this type of monotonicity (a large proportion of respondents failing to choose the dominant option could be considered to be a sign of poor data quality). A balance of choice types across the blocks was sought. For example, all of the blocks contained at least one (but no more than two) choice sets of type 1. Apart from this manual distribution of choice types, the choice sets were assigned to blocks at random.

When asked to choose between multiple options laid out next to each other, it is possible that a 'left-hand-side' bias may exist if respondents (subconsciously or otherwise) treat the option on the left as the default choice (Spalek and Hammad, 2005). Similarly, a 'top-to-bottom' bias may exist when information or options are laid out one on top of the other (Mulhern *et al.*, 2016). To control for potential bias due to the positioning of options, eight 'mirror' blocks were generated to match the eight blocks mentioned above. These consisted of the same 10 choice sets but switched the labels assigned to the two alternatives – i.e. the alternative labelled as

'patient A' in the original block choice set appeared as 'patient B' in the corresponding mirror block choice set (and *vice versa*). Including these mirror blocks meant that there were a total of 16 different versions of the survey.

5.2.1.4 Extension tasks

As mentioned above, extension choice sets were included at the end of each block to examine whether respondents' choices are influenced by information about how long the patients have known about their illness. Each extension choice set replicated the scenario depicted in one of the DCE choice sets, but adding information that one of the patients had known about their illness for two years whereas the other patient had just found out about their illness. An example is shown in Table 5-2.

Table 5-2. Example of standard and corresponding extension choice set

	Standard DCE choice set		Corresponding extension choice set	
Attribute	Patient A	Patient B	Patient A	Patient B
Life expectancy without treatment	12 months	3 months	12 months	3 months
Quality of life without treatment	50%	50%	50%	50%
Life expectancy gain from treatment	1 month	6 months	1 month	6 months
Quality of life gain from treatment	25%	25%	25%	25%
How long patient has known about illness	No information provided		0 years (just found out)	2 years

In the standard DCE choice set, patient B is in poorer health than patient A without treatment (three months of life expectancy at 50% quality of life versus 12 months of life expectancy at 50% quality of life). Choosing to treat patient B would be consistent with a preference for giving priority to those who are worse off without treatment. In the extension choice set, the respondent is told that patient B has known about their illness for two years whereas patient A has only just learned of their illness. Some respondents who chose to treat patient B in the standard DCE choice set may have done so because of a concern about how little time they have to 'get their affairs in order'. If so, they may switch to choosing to treat patient A in the extension choice set, as patient A will have had less time to prepare than patient B when taking into account the time with knowledge of their prognosis.

Eight standard DCE choices sets were hand-picked to form the basis for the extension tasks. The selection was guided by judgements about whether the choice sets depicted scenarios of particular interest (such as the example shown in Table 5-2) and by considerations about whether they could be presented graphically using a format similar to the one used for the standard DCE choice sets.

Two extension choice sets were included in each block and presented to respondents after they had completed the 10 standard DCE choice sets. One of the extension choice sets replicated a standard DCE choice set that respondents in that block would have already completed, to allow within-respondent comparisons; the other replicated a standard DCE choice set from a different block. The latter was always presented first, so respondents were never faced with an extension choice set immediately following the standard DCE choice set upon which the extension choice set had been based. For every choice set in which the time with knowledge was given to one of the patients, there was corresponding choice set (in a different block) which was identical except that the time with knowledge was given to the other patient. As with the standard DCE choice sets, mirror choice sets were used in an attempt to control for potential top-to-bottom bias.

5.2.1.5 Presentation of choice tasks

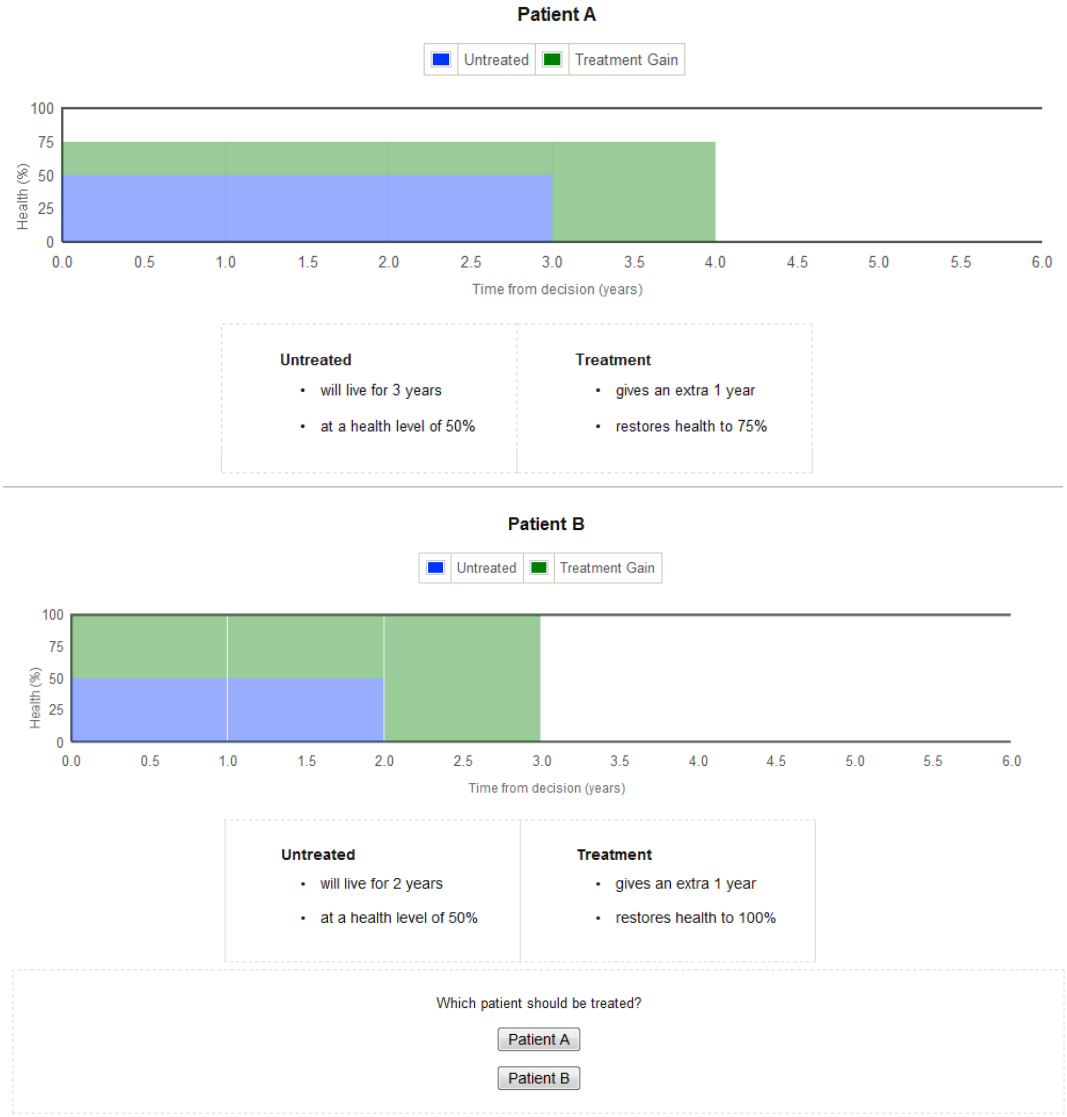
Following empirical studies 1 and 2, the attributes and levels were presented as characteristics of two hypothetical patients (patient A and patient B) and the effects of the treatments available to them. Adapting the design of an existing survey used in research elsewhere (Rowen *et al.*, 2016a), the information was presented using a combination of diagrams and text descriptions (Figure 5-1). The diagrams used a horizontal scale to represent life expectancy and a vertical scale to represent quality of life (described in the survey as 'health'). They appeared directly above the corresponding text descriptions, which were presented using bullet points. The use of both diagrams and text to present the choice set information was informed by feedback provided by respondents in empirical study 1 (see 3.3.2). Due to space restrictions, the diagrams and text for patient A always appeared directly above the diagrams and text for patient B, with the choice options at the bottom.

The survey began with instructions (reproduced in full in Appendix 8) introducing the diagrams as a way of showing how different illnesses and treatments affect people's health and life expectancy. Respondents were asked which patient they thought should be treated, assuming that the health service had only enough funds to treat one of the two patients, and that there were no alternative treatments available. It was emphasised that there were no right or wrong answers.

Respondents were advised that they would be given information about the patients' health and life expectancy with and without treatment, but that no other information about the patients was available (except that they were both adults). To prevent respondents from making choices based on hope that a cure for the illnesses may be found in the future, they were told that 'the nature of the illnesses is such that further treatment will not be possible if either patient is not treated

today – this is the only opportunity for treatment.' Although such 'hope effects' may exist and influence people's choices regarding priority-setting, they were not considered pertinent to this study. The treatments typically considered under NICE's end of life policy tend not to offer life extensions that are long enough for the realistic possibility of cures being discovered and made available for use during the intervening period.

Figure 5-1. Example of DCE task diagram and text



No indifference or 'status quo' option was offered. The use of questions involving forced choices without an indifference option is a departure from the approach used in empirical studies 1 and 2, and was informed by a number of considerations. First, it was felt that even if respondents found it difficult to choose between the two patients, they would nevertheless prefer to treat one of them rather than to treat neither, since some health gain is preferable to the baseline of no health gain to either. Second, it was suspected that such an option may be used as a default ('opt-out') choice, thus providing a way for respondents to avoid taking time to

make difficult decisions. This was a particular concern due to the unsupervised, self-complete survey setting (see 5.2.2), which may encourage respondents to seek shortcuts in order to complete the survey as quickly as possible. Bridges *et al.* (2011) advise that the inclusion of indifference options results in the censoring of data, which can limit researchers' ability to estimate the underlying preference structure. Finally, if respondents are genuinely indifferent between treating the two patients, this should result in a roughly even split between patient A and patient B in the choice data. The use of mirror choice sets controls for the possibility that respondents will revert to a default choice, such as the patient presented first, every time they are unable to choose between the patients.

The 10 standard DCE tasks were presented to respondents in a random order so as to ensure that order bias was not systematic across the sample. After the standard tasks, further instructions were provided to explain the additional 'time with knowledge' attribute. The diagrams were modified to incorporate this attribute (Figure 5-2 and Figure 5-3). Respondents were again asked to indicate which patient they thought should be treated. After completing the extension tasks, respondents were asked background questions. Finally, they were invited to leave feedback using an open-ended comment box. The survey allowed respondents to go back to previous questions and change their answers if they so wished.

Figure 5-2. Extension task diagram example (time with knowledge = 2 years)

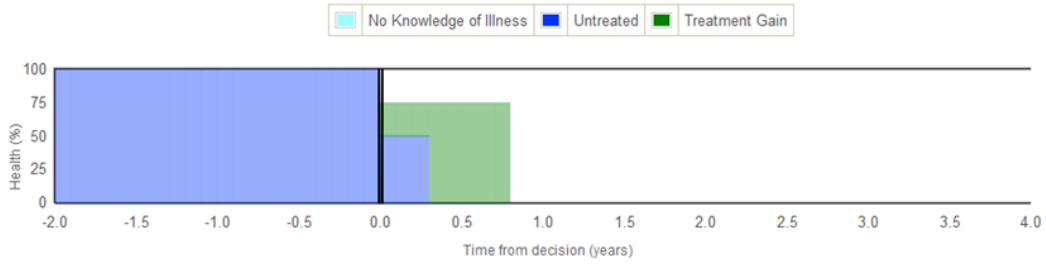
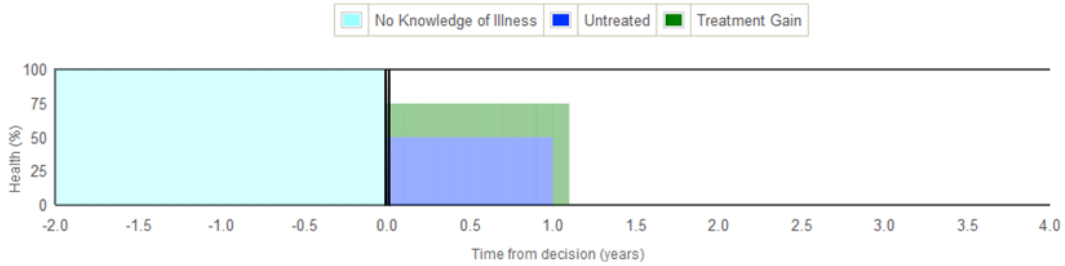


Figure 5-3. Extension task diagram example (time with knowledge = 0 years)



5.2.2 Administration of survey

The choice sets were included in a self-completion survey administered over the internet. The survey was developed in collaboration with epiGenesys, a software development company.

Internet surveys offer a quick and cost-effective means of collecting a large amount of choice data. Large samples are difficult to achieve using other modes of administration: postal self-complete surveys have very low responses rates; surveys administered as part of face-to-face interviews are expensive to manage; and the complexity of the questions in this study precluded the use of telephone-based data collection. By comparison, internet surveys can be custom-designed to present information and to collect, store and export data in a clear, user-friendly manner.

Interviewer-led survey administration is often preferred because the interviewer can explain the instructions more fully if required (Bridges *et al.*, 2011), and respondents may be more likely to give their full attention to the survey whilst being guided (Shah *et al.*, 2013b). However, the use of interviewers can lead to forms of interviewer bias – for example, if when explaining the instructions the interviewer gives subtle clues that influence the respondent towards certain preferences or choice strategies. With internet surveys, the questions and instructions are presented in the same manner to all respondents (although presentation may differ according to the hardware or software being used, it is reasonable to assume that any variability will be random and unlikely therefore to result in systematic bias).

The vast majority of households in the UK now have access to the internet (Office for National Statistics, 2016), but there remain concerns about the extent to which a sample of online panel members can be representative of the general population. Although quotas can be used to ensure representativeness in terms of certain observable characteristics (such as age), it is likely that the sample will still be systematically different in terms of other unobservable characteristics. However, this issue is not specific to web-based data collection. The types of individuals who are willing to complete postal surveys or to allow interviewers into their homes for face-to-face interviews, for example, are similarly unlikely to be representative of the general population. Some market research agencies advise that providing an incentive for completing surveys can help to improve representativeness as an unpaid survey is more likely to be completed only by those passionately interested in the subject of that particular survey (YouGov, 2017).

5.2.3 Sample

The survey was administered on a sample of adult members of the general public in England and Wales, all of whom were members of a panel of a market research agency, ResearchNow. A 'minimum quota' approach, combined with a targeted invitation strategy, was used to ensure that the sample was broadly representative of the general population in terms of key observable characteristics. The target sample size of 4,000 was determined on the basis that this was the largest sample that could be recruited within the required timelines. Individuals who had recently completed health-related surveys were not invited to take part. The average panel member completes six surveys per year. Completion statistics, including the age, gender and social grade of respondents who had completed the survey, were checked daily and used to guide the targeting of invitations. Once a quota for a particular subgroup had been reached, individuals attempting to access the survey who fell within that subgroup were 'screened out' and informed that they were not eligible to take part. Once respondents had been 'screened in' and given their informed consent to take part, they were randomly assigned to one of the 16 blocks. Respondents were compensated by way of 'reward points' which can be redeemed for gift vouchers or charity donations.

5.2.4 Ethical approval

The survey design 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. The ethics approval required the destruction of any information provided by respondents who did not complete the survey in full.

5.2.5 Piloting

The main study was preceded by a pilot, which used a convenience sample of 12 members of non-academic staff and postgraduate research students at the University of Sheffield (excluding those in the Faculty of Medicine, Dentistry and Health or the Department of Economics). The pilot comprised face-to-face interviews conducted by author in which respondents completed the survey (accessed via a laptop connected to the internet) without assistance, and then answered probing questions designed to elicit feedback and concerns about the survey and approach.

The pilot was completed successfully, supporting the acceptability of the text and diagrams used in the survey and the feasibility of the proposed methods (choice of

attribute levels, forced choices, web-based survey, randomisation processes). The instructions and choice tasks were described by most of the pilot respondents as being clear and easy to follow. Some of the wording of the instructions was improved following feedback from two of the respondents. All of the respondents stated that they were able to understand and complete the questions without assistance. The levels of understanding and engagement (as perceived by the interviewer) were high. Respondents spent between six and 14 minutes completing the questions (mean: 9 minutes and 10 seconds).

Further testing was conducted by way of a 'soft launch' data collection strategy. Once approximately 750 respondents had completed the survey, the survey was closed and the data were checked for issues. Whilst the average time taken to complete the questions (choice tasks and follow-up questions) was consistent with the pilot (mean: 9 minutes and 44 seconds), it was noted that 14 respondents (2%) completed the questions in less than three minutes. It was questioned whether it was possible to complete the survey this quickly whilst paying adequate attention to the tasks at hand. When faced with choice sets in which one alternative dominated the other, nine of these 14 respondents (64%) failed to choose the dominant alternative. Since patient A and patient B were overall equally likely to be represent the dominant alternative in these choice sets, a respondent who is not taking the survey seriously (for example, making choices at random) is expected to have a 50% chance of choosing the dominant alternative. By comparison, only 12% of respondents who spent at least three minutes completing the questions failed to choose the dominant alternative. It was therefore deemed reasonable to exclude from the analysis data for respondents who completed the questions in less than three minutes on grounds of poor data quality.

The soft launch approach also provided an opportunity to examine the open-ended comments left by respondents, in case these highlighted any problems with the survey. Of the 100 or so comments that had been left, the majority were positive (for example: "very well set out and easy to navigate"). Three respondents left comments about one of the background questions, stating that they were unsure about which category they belonged to when asked about the occupation of the chief income earner of their household. The instructions to the question were amended to address these comments. No other changes to the survey were deemed necessary in light of the soft launch data analysis.

5.2.6 Methods of analysis

Descriptive analyses were used to determine the 'level of agreement' amongst respondents (the proportion choosing the majority choice) for each choice set. The

average level of agreement for each choice type (see 5.2.1.3) was then calculated by grouping choice sets according to the nature of the choices depicted.

The choice data were modelled using a random utility maximisation framework (Louviere *et al.*, 2000) and the Stata software package. As the data were binary choice data – 1 representing one option being chosen and 0 representing the other being chosen – conditional logit regressions were used.

The model estimated is of the form:

$$V = \beta_1 LE\ without\ treatment + \beta_2 QOL\ without\ treatment$$
$$+\beta_3(QOL\ gain * LE\ without\ treatment)$$
$$+\beta_4(LE\ gain * QOL\ without\ treatment)$$
$$+\beta_5(LE\ gain * QOL\ gain)$$
$$+\beta_6 EOL$$

The deterministic component of the utility function (*V*) is a function of the attribute levels between alternatives, where the coefficients β_1 to β_6 are estimated in the model. The explanatory variables *LE* (life expectancy) without treatment and *QOL* (quality of life) without treatment represent the baseline health of the patients. The three interactions terms together make up QALY gains (quality of life improvement for a given level of life expectancy; life extension for a given level of quality of life; and life extension combined with quality of life improvement). These variables were treated as continuous. An end of life dummy variable was also included. This took a value of 1 for profiles that would meet the NICE criteria for defining a life-extending end of life treatment (i.e. life expectancy without treatment of less than or equal to 24 months; life expectancy gain of greater than or equal to three months) and 0 otherwise.[19] The performance of models both with and without the end of life dummy was assessed by examining the Akaike and Bayesian information criteria (Akaike, 1973; Schwartz, 1978) and conducting likelihood ratio tests.

The coefficients estimated in the model can be summed to give the overall utility for each profile (combination of attribute levels). This gives an indication of the relative social value of the 110 profiles in the experimental design. As described in 5.2.1.1, the probability of choosing a given profile from the complete set of profiles can be predicted from the model estimates. Following the approach used by Green and Gerard (2009), the relative predicted probabilities for all of the 110 profiles were calculated, allowing comparisons between the profiles with higher probabilities

[19] An alternative model, in which the end of life dummy was defined in terms of life expectancy without treatment but not life expectancy gain, was also tested.

(those which are likely to be most preferred overall) with those with lower probabilities (those which are likely to be least preferred overall).

Finally, a selection of respondent subgroups whose choices may be expected to differ from those of the rest of the sample were defined *a priori*. These were: (1) respondents with experience of close friends or family with terminal illness; (2) respondents with responsibility for children aged under 18 years; (3) respondents who opted to leave a comment in the open-ended box at the end of the survey (this was not mandatory); and (4) respondents who completed the questions much quicker than average. Family circumstances and personal experience of terminal illness were mentioned as influences on respondents' preferences in empirical study 1 (see 3.3.2); whether a comment was left and how quickly the questions were completed may be indicators of respondent engagement. For each subgroup, the best fitting model was estimated and the results were compared to those of the same model using the full sample.

Throughout the analyses a zero discount rate was assumed based on the lack of evidence from empirical studies 1 and 2 that time preference is a motivating factor for giving higher priority to the treatment of end of life patients.

5.3 Results

Data collection was undertaken in early 2012. In total, 43,000 individuals were invited by email to take part in the survey, of whom 5,308 clicked on the link to access the survey (response rate: 12.3%). Of the individuals who accessed the survey, 4,008 completed the survey in full (completion rate: 75.5%). The remainder either did not give consent to take part, or began the survey but dropped out without completing all of the questions. The response and completion rates for this survey are consistent with those of similar internet surveys whose sample comprised members of ResearchNow's panel.

As mentioned above (see 5.2.5), it was agreed that data for respondents who spent less than three minutes on the questions would be suppressed from the final data set. This cut-off excluded 39 respondents, leaving a sample of 3,969 respondents (47,628 pairwise observations). Of these 47,628 observations, 39,690 were for the standard DCE tasks to be analysed using the conditional logit model; the remaining 7,938 were for the extension tasks.

The background characteristics of the sample are presented in Table 5-3. By design, the sample was broadly representative of the general population with respect to age and gender (Office for National Statistics, 2011). Despite the use of quotas to seek representativeness in terms of social grade, the sample comprised a

slightly larger proportion of individuals in the highest and very lowest grades than in the general population (National Readership Survey, 2012-3), presumably due to changes in circumstances since these individuals joined the panel.

Table 5-3. Sample background characteristics

		n	%	Population
Total		3,969	100.0%	
Age (years)	18-29	730	18.4%	21%
	30-44	1,087	27.3%	26%
	45-59	855	21.5%	25%
	60+	1,297	32.7%	28%
Gender	Female	2,027	51.1%	51%
	Male	1,942	48.9%	49%
Social grade [a]	A	221	5.6%	4%
	B	1,114	28.1%	22%
	C1	1,150	29.0%	29%
	C2	645	16.3%	21%
	D	357	9.0%	15%
	E	482	12.1%	8%
Household composition	Responsible for children	963	24.3%	
	Not responsible for children	3,006	75.7%	
Education	None beyond minimum school leaving age	889	22.4%	
	Beyond minimum school leaving age; no degree	1,244	31.3%	
	Beyond minimum school leaving age; degree	1,836	46.3%	
Self-reported general health	Very good	1,008	25.4%	
	Good	1,958	49.3%	
	Fair	770	19.4%	
	Poor	210	5.3%	
	Very poor	23	0.6%	
Experience of close friends or family with terminal illness	Yes	2,689	67.8%	
	No	1,197	30.2%	
	Question skipped by respondent	83	2.1%	

[a] Refers to the occupation/qualifications/responsibilities of the chief wage earner of the respondent's household; see National Readership Survey (2012-13).

Three hundred and eighty-nine respondents (9.8%) failed to choose the dominant option when faced with choice sets in which one alternative dominated the other (i.e. where both patients have the same amount of life expectancy and quality of life without treatment, but one patient gains more life expectancy and more quality of life from treatment than the other). However, it is not necessarily the case that these preferences are 'irrational' – Lancsar and Louviere (2006) warn against researchers imposing their own preferences by deleting responses that do not conform to their expectations. Data for these respondents were therefore included in the analysis.

5.3.1 Descriptive statistics of the choices made

Table 5-4 reports the average level of agreement for the choice sets belonging to each choice type (see 5.2.1.3). The majority of respondents chose to treat the patient who gains more from treatment, regardless of whether that patient is better or worse off without treatment. This is demonstrated by the high levels of agreement for choice sets corresponding to choices types 2, 13 and 6. Across the three choice sets in which the gains from treatment are the same for both patients and one patient is worse off without treatment in terms of both life expectancy and quality of life (choice type 10), the better off patient was chosen 66% of the time.

Table 5-4. Average level of agreement, by choice type

Choice type	No. choice sets	Description	Level of agreement (% respondents who chose patient X)
2	11	Both patients have the same LE / QOL without treatment. Patient X gains more LE and more QOL from treatment than patient Y.	92%
13	5	Patient X has shorter LE and higher QOL without treatment and gains more LE from treatment than patient Y.	85%
6	1	Patient X has lower QOL without treatment and gains more LE and more QOL from treatment than patient Y.	85%
1	14	Patient X has longer LE without treatment and gains more QOL from treatment than patient Y.	78%
12	2	Patient X has shorter LE without treatment and gains more LE from treatment than patient Y.	76%
9	4	Patient X has longer LE without treatment and gains more LE from treatment than patient Y.	74%
11	2	Patient X has longer LE and lower QOL without treatment and gains more QOL from treatment than patient Y.	72%
8	4	Patient X has shorter LE without treatment and higher QOL without treatment than patient Y. Both patients gain same amount of LE / QOL from treatment.	68%
7	5	Patient X has shorter LE and lower QOL without treatment and gains more QOL from treatment than patient Y.	68%
10	3	Patient X has longer LE and higher QOL without treatment than patient Y. Both patients gain same amount of LE / QOL from treatment.	66%
3	10	Patient X has shorter LE without treatment and gains more QOL from treatment than patient Y.	62%
4	10	Both patients have the same LE / QOL without treatment. Patient X gains more QOL from treatment; patient Y gains more LE from treatment.	59%
5	9	Patient X has lower QOL without treatment and gains more QOL and less LE from treatment than patient Y.	58%

Overall, there was a statistically significant tendency (Student's t-test; $p<0.01$) to choose to treat the alternative labelled patient B (the alternative appearing at the

bottom of the respondent's screen). Examining differences between mirror blocks, wherever the most popular choice was patient A (B) in the original block, the most popular choice was always patient B (A) in the corresponding mirror block.

5.3.2 Discrete choice model results

Table 5-5 reports the results of the conditional logit modelling. Note that the parameters have been coded such that that one year in full health is given a value of 1. The results of two models are presented – one with and one without the end of life dummy variable described in 5.2.6. The model *with* the end of life dummy performed better according to the Akaike and Bayesian information criteria and likelihood ratio tests ($p<0.01$).

Table 5-5. Conditional logit modelling results

	Model without end of life dummy [a]			Best fitting model (*with* end of life dummy) [b]		
Attribute	Coefficient	Std. error	p-value	Coefficient	Std. error	p-value
LE without treatment	-0.10715	0.00696	0.00	-0.06945	0.00736	0.00
QOL without treatment	-0.06357	0.04877	0.19	0.00051	0.04936	0.99
Interaction: QOL gain # LE without treatment	0.81567	0.01652	0.00	0.84535	0.01682	0.00
Interaction: LE gain # QOL without treatment	2.71342	0.05990	0.00	2.39408	0.06305	0.00
Interaction: LE gain # QOL gain	3.17557	0.10330	0.00	2.76204	0.10616	0.00
End of life dummy	N/A	N/A	N/A	0.37253	0.02510	0.00

[a] Akaike information criterion = 43577; Bayesian information criterion = 43623
[b] Akaike information criterion = 43358; Bayesian information criterion = 43414

In both models, the coefficient for life expectancy without treatment is negative and statistically significant, which indicates that respondents were more likely to choose to treat the patient with shorter life expectancy without treatment, *ceteris paribus*. The coefficient for quality of life without treatment is not statistically significant in either model. The coefficients for the three interactions that make up QALY gains are all positive and statistically significant, and considerably larger in magnitude than the coefficient for life expectancy without treatment. The coefficient for the interaction between life expectancy gain and quality of life without treatment is substantially larger than the coefficient for the interaction between quality of life gain and life expectancy without treatment. This indicates that respondents' choices are driven by life extensions to a greater degree than by quality of life improvements. The coefficient for the end of life dummy is positive and statistically

significant, which indicates that respondents are more likely to choose a treatment that meets the NICE criteria than one that does not.

To assist interpretation of the model results, Table 5-6 presents the utility scores based on the best fitting model for a selection of the profiles, as well as the predicted probability of choosing each profile from the full set of 110 profiles. A table showing the same information for all 110 profiles can be found in Appendix 12.

Table 5-6. Estimated utility score and probability of choice for the highest/lowest ranked profiles

Rank	LE without treatment (mths)	QOL without treatment (%)	LE gain (mths)	QOL gain (%)	QALYs without treatment	QALYs gained	Utility	Prob.	Cumul. Prob.
1	60	50	12	50	2.500	3.500	4.3445	0.1351	0.1351
2	36	50	12	50	1.500	2.500	3.6380	0.0667	0.2018
3	12	50	12	50	0.500	1.500	3.3041	0.0477	0.2495
4	24	50	12	50	1.000	2.000	3.2848	0.0468	0.2964
5	60	50	6	50	2.500	3.000	3.0554	0.0372	0.3336
6	3	50	12	50	0.125	1.125	3.0392	0.0366	0.3702
7	3	100	12	0	0.250	1.000	2.7498	0.0274	0.3976
8	12	100	12	0	1.000	1.000	2.6977	0.0260	0.4237
9	60	50	12	25	2.500	2.000	2.5973	0.0235	0.4472
10	60	50	3	50	2.500	2.750	2.4109	0.0195	0.4668
11	12	50	12	25	0.500	1.000	2.4022	0.0194	0.4861
12	36	50	6	50	1.500	2.000	2.3490	0.0184	0.5045
13	36	50	12	25	1.500	1.500	2.3135	0.0177	0.5222
14	3	50	12	25	0.125	0.813	2.2958	0.0174	0.5396
15	24	100	12	0	2.000	1.000	2.2557	0.0167	0.5564
16	60	50	2	50	2.500	2.667	2.1961	0.0158	0.5721
17	36	100	12	0	3.000	1.000	2.1862	0.0156	0.5878
18	24	50	12	25	1.000	1.250	2.1716	0.0154	0.6031
19	60	100	12	0	5.000	1.000	2.0474	0.0136	0.6167
20	12	50	6	50	0.500	1.000	2.0150	0.0132	0.6299
-	-	-	-	-	-	-	-	-	-
10 most preferred profiles	2.750	0.600	0.875	0.375	1.438	2.038	3.1121	0.04668	
20 most preferred profiles	2.638	0.625	0.846	0.313	1.600	1.680	2.6677	0.03149	
55 most preferred profiles	2.268	0.600	0.558	0.277	1.305	1.201	1.7856	0.01570	
55 least preferred profiles	2.232	0.627	0.170	0.132	1.457	0.310	0.3081	0.00249	
20 least preferred profiles	2.488	0.625	0.117	0.050	1.644	0.093	0.0156	0.00179	

Rank	LE without treatment (mths)	QOL without treatment (%)	LE gain (mths)	QOL gain (%)	QALYs without treatment	QALYs gained	Utility	Prob.	Cumul. Prob.
10 least preferred profiles	3.225	0.600	0.108	0.025	2.013	0.069	-0.0687	0.00164	
-	-	-	-	-	-	-	-	-	-
91	24	50	3	0	1.000	0.125	0.1606	0.0021	0.9662
92	12	50	0	25	0.500	0.250	0.1421	0.0020	0.9683
93	12	100	1	0	1.000	0.083	0.1306	0.0020	0.9703
94	12	50	2	0	0.500	0.083	0.1303	0.0020	0.9723
95	36	50	3	0	1.500	0.125	0.0912	0.0019	0.9742
96	3	50	0	50	0.125	0.125	0.0886	0.0019	0.9761
97	3	50	1	0	0.125	0.042	0.0826	0.0019	0.9780
98	24	100	1	0	2.000	0.083	0.0611	0.0019	0.9799
99	24	50	2	0	1.000	0.083	0.0609	0.0019	0.9817
100	60	100	2	0	5.000	0.167	0.0523	0.0018	0.9836
101	3	50	0	25	0.125	0.063	0.0357	0.0018	0.9854
102	12	50	1	0	0.500	0.042	0.0306	0.0018	0.9872
103	36	100	1	0	3.000	0.083	-0.0083	0.0017	0.9889
104	36	50	2	0	1.500	0.083	-0.0086	0.0017	0.9907
105	24	50	1	0	1.000	0.042	-0.0389	0.0017	0.9924
106	60	50	3	0	2.500	0.125	-0.0477	0.0017	0.9940
107	36	50	1	0	1.500	0.042	-0.1083	0.0016	0.9956
108	60	100	1	0	5.000	0.083	-0.1472	0.0015	0.9971
109	60	50	2	0	2.500	0.083	-0.1475	0.0015	0.9986
110	60	50	1	0	2.500	0.042	-0.2472	0.0014	1.0000

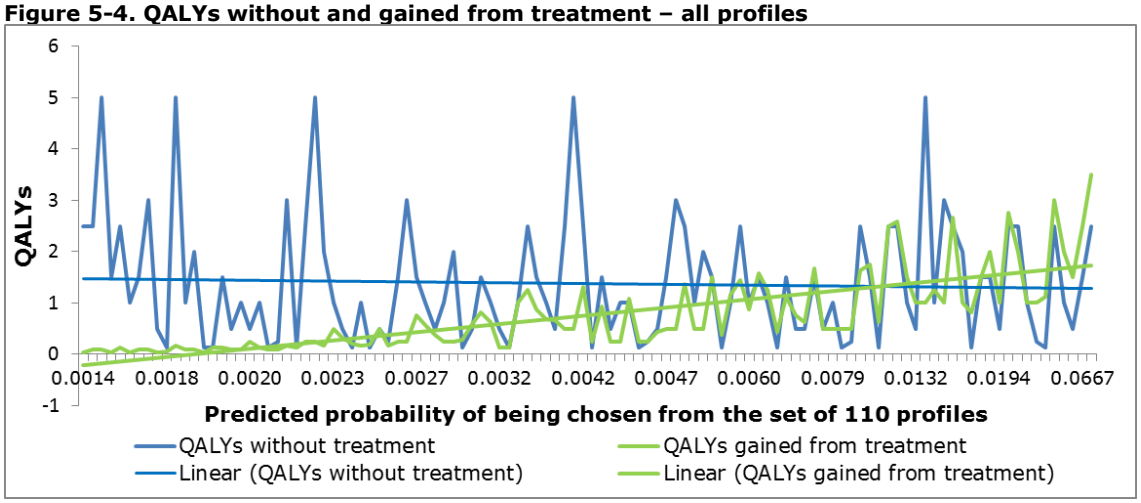
Note: all outcomes are undiscounted

The highest ranked profiles all involve substantial treatment gains. All of the profiles ranked between 1st and 25th involve a life expectancy gain of 12 months and/or a quality of life gain of 50%. By contrast, the lowest ranked profiles mostly involve a small life expectancy gain and no quality of life gain. A similar pattern with respect to life expectancy without treatment does not exist – profiles involving the highest and lowest levels for this attribute (60 months and three months, respectively) appear at both the top and the bottom of Table 5-6. Quality of life without treatment is 50% in most of the highest ranked profiles, but this is always accompanied by a non-zero quality of life gain from treatment. There is little difference between the highest and lowest ranked profiles in terms of QALYs without treatment – the key driver is the difference in the sizes of the QALY gains from treatment.

Figure 5-4 illustrates the levels of QALYs without treatment and QALYs gained from treatment associated with all of the 110 profiles, where the horizontal axis represents the standardised predicted probabilities from the lowest (least preferred)

to the highest (most preferred) profile. Whilst the patterns are noisy, the green linear trend line for QALYs gained from treatment has a clear upward slope (the larger the size of the QALY gains, the greater the probability of the profile being chosen). The blue linear trend line for QALYs without treatment is relatively flat, indicating that the number of QALYs without treatment does not have a major effect on the probability of the profile being chosen.

Figure 5-4. QALYs without and gained from treatment – all profiles



5.3.3 Subgroup analysis

As described in 5.2.6, respondents were assigned to subgroups according to their responses to the background questions (whether or not they have experience of terminal illness in close friends or family; whether or not they have children) or to the ways in which they completed the survey (whether they left a comment or not; how quickly they completed the survey). The best fitting models were estimated for each subgroup and the results were compared to those of the same model using the full sample. This analysis indicated no difference in the signs or approximate magnitude of the coefficients for any of the subgroups compared with the entire sample (except for the coefficient for quality of life without treatment, which was not robust across models and never found to be statistically significant).

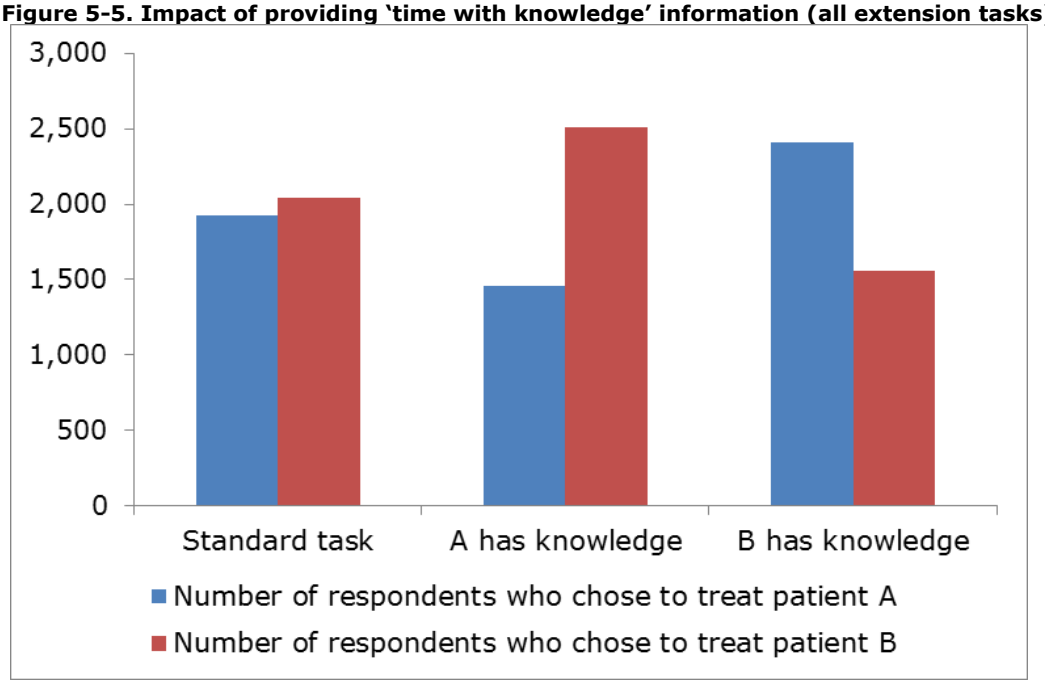
The best fitting model was also run excluding the 389 respondents who failed to select the dominant alternative when faced with choice sets in which one alternative dominated the other. Excluding these respondents did not affect the conclusions from the regression results.

5.3.4 Extension tasks

Comparing the response data for the extension tasks with the data for the corresponding standard DCE tasks allows the testing of whether respondents are more likely to choose to treat the patient who has just found out about their illness

(for example, due to concerns about how long they have to prepare for death). In all of the 16 extension tasks, being told that one of the patients has known about their illness for two years increased the proportion of respondents choosing to treat the other patient compared to when no 'time with knowledge' information is provided. In six of the 16 cases, this increase was sufficiently large that the majority choice in the extension task flipped from the majority choice in the corresponding standard task. Figure 5-5 presents the impact on choices of providing information on how long the patients have known about their illness, summed across all 16 extension tasks.

Figure 5-5. Impact of providing 'time with knowledge' information (all extension tasks)



The choice set that formed the basis for the example in Table 5-2 was identified during the study design phase as being of particular interest. Quality of life (both before and after treatment) is the same for both patients. One patient has shorter life expectancy without treatment than the other (3 months < 12 months), and despite gaining more life expectancy from treatment (6 months > 1 month), that patient continues to have shorter life expectancy *after* being treated (9 months < 13 months). If people wish to give priority to those with shorter life expectancy and prefer larger health gains to smaller health gains, then it is expected that most people would choose to treat this 'worse off, larger gain' patient. This is indeed the case in the two standard DCE tasks that mirror each other (75% and 78% of respondents chose to treat this patient).

In two of the extension tasks based on this choice set (and its mirror), respondents were told that the 'worse off, larger gain' patient found out about their illness two years ago, while the other ('better off, smaller gain') patient had only just found

out about their illness. This means that despite their shorter life expectancy, the 'worse off, larger gain' patient will have had longer to prepare for death (25 months > 12 months). In light of this new information, a smaller proportion, but still the majority, of respondents chose to treat this patient (57% and 57%).

5.4 Discussion

This study used a web-based DCE to elicit the preferences of a large general public sample in England and Wales over a range of health care priority-setting scenarios, focusing on social preferences regarding the prioritisation of treatments for patients with short life expectancy. The results show that choices about which patient to treat are influenced more by the sizes of the gains achievable from treatment than by patients' life expectancy or quality of life in absence of treatment. The profiles most likely to be chosen were those with the highest levels of both life expectancy gain (12 months) and quality of life gain (50%). Likewise, the profiles least likely to be chosen were those with very small gains. On the other hand, the data suggest that the level of life expectancy without treatment in a given profile has little impact on the likelihood of that profile being chosen. There is certainly no indication that being at the end of life is the driving factor; in fact, the average level of life expectancy without treatment in the 55 profiles most likely to be chosen is almost identical to that in the 55 profiles that are least likely to be chosen (Table 5-6).

Analysis at the individual choice set level confirms this: in several of the choice sets showing the highest levels of agreement amongst respondents (Table 5-4), the most popular choice was to treat the patient with longer left to live and for whom treatment offered larger health gains, in favour of the patient with shorter life expectancy. The overall view seems to be that giving priority to those who are worse off is desirable, but only if the gains from treatment are substantial.

The results show that people's preferences are heterogeneous. Although the conditional logit model did not account for the panel nature of the data, the analysis of choice frequencies at the individual respondent level showed that some respondents appeared to support a QALY-maximisation type objective throughout; a small minority always sought to treat those who are worse off without treatment; but the majority seemed to advocate a mixture of the two approaches. These heterogeneous preferences do not appear to be well predicted by respondents' observable characteristics (see Appendix 13).

The finding of this study that respondents attach relatively little weight to how much life expectancy (and quality of life) without treatment patients have does not necessarily refute evidence elsewhere in the literature of popular support for the

use of severity as a priority-setting criterion (Shah, 2009). This study focused on a small range of scenarios, all of which involved relatively poor prognoses (in terms of life expectancy). Across all of the profiles included in the design, the patient who is 'best off' without treatment would still die within five years. Thus, in effect, *all* of the profiles in the study describe patients who are at or near their end of life to some extent. It is not possible from these data alone to infer whether the importance of the life expectancy without treatment attribute would be markedly different in a survey asking respondents to choose between treating patients with very short life expectancies and treating patients with much longer life expectancies (for example, patients with 30 years of remaining life, who clearly cannot be described as 'end of life').

The outcomes examined in this study were not adjusted to account for any possible social time preference. Applying a positive discount rate would likely further strengthen the finding that respondents do not place special value on treating patients with short life expectancy, though the effect of discounting is expected to be quite small given the relatively short timeframes included in the study design.

The internet survey provided an efficient means of obtaining a large sample. The response rate observed is not unusual for a non-probability-based panel sample, and cannot easily be compared with response rates from studies using different modes of administration (for example, because many of the individuals invited to take part may not be active members of the panel) (Baker *et al.*, 2010b). However, this mode of administration offers limited opportunity for debriefing with respondents about their experience of completing the survey (although the earlier studies and piloting were useful in this respect). The study was designed in such a way that the ranking of the profiles would not be expected to differ if some respondents failed to pay adequate attention to the choice tasks (for example, making choices at random). Nevertheless, if respondents had failed to understand the instructions, then this could be problematic. For example, they may mistakenly believe that the tasks require them to choose which patient they would prefer to be in the position of, rather than which patient they would prefer the health service to treat. A useful addition to future stated preference studies, particularly those administered in an unsupervised setting, would be to design follow-up questions that can be used to check whether respondents agree with the policy implications of their responses to the DCE questions. For example, asking respondents to rank a variety of statements describing different priority-setting approaches according to the extent to which they agreed with them would provide information about which broad priority-setting objectives they find most and least acceptable. A more explicit method would be to present respondents with a statement such as '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' and ask them to indicate whether they agreed or disagreed with that statement. This would allow the checking of whether respondents agree with a policy statement that appears to match their responses in the DCE tasks. A high level of agreement would add legitimacy to the DCE results.

The finding that respondents were more likely to choose the alternative appearing at the bottom of the screen is consistent with findings reported elsewhere in the literature on bias due to the positioning of choice options (Spalek and Hammad, 2005). The use of randomisation procedures and mirror choice sets to minimise the impact of ordering-related biases is recommended.

Results from the extension tasks show that including information about the amount of time that patients have known about their prognosis has a clear impact on preferences – specifically, holding all else constant, respondents are less likely to choose to treat a patient if that patient has known about their illness for two years than if they have only just found out about their illness. This suggests that the observed tendency to give priority to the end of life patient may be driven by concerns about the patient's ability to prepare for death rather than the amount of time they have left to live *per se*. The fact that this time with knowledge attribute was clearly the main subject of the extension task instructions and questions (see Appendix 9) is likely to have resulted in a focusing effect whereby respondents placed more importance on this attribute than they otherwise might have done. Furthermore, the extension tasks in this study did not allow for the elicitation of the strength of respondents' preferences. Further investigation of preferences regarding preparedness is recommended.

NICE's current criteria for determining whether a treatment should be a candidate for special consideration are that it is indicated for patients with less than 24 months of life expectancy and that it extends life by at least three months. Hence, a treatment offering 0.5 QALYs through a 12 month life expectancy gain (and no quality of life gain) to patients with 24 months life expectancy at 50% quality of life without treatment would meet these criteria. An alternative treatment, also offering 0.5 QALYs through a 25% quality of life gain (and no life expectancy gain) to the same patients would *not* meet the criteria for being eligible for special consideration. The results of this study indicate that the profile representing the former treatment would in fact be more likely to be chosen (ranked 44th with a 0.51% probability of being chosen) than the profile representing the latter treatment (ranked 83rd; 0.23%) (Appendix 12). This suggests that the focus on life

extensions and absence of quality of life improvements in the criteria may be consistent with public preferences, although some of the descriptive statistics analysis (Table 5-4) suggests otherwise.

An examination of the impact of marginal changes in any of the attribute levels from a profile representing a treatment that just meets the current NICE end of life criteria suggests that amending the life expectancy without treatment criterion would not have a major effect on utility. The predicted probability of choosing a profile involving a life expectancy gain of three months is much the same regardless of whether the patient's life expectancy without treatment is three, 24 or 36 months. By comparison, a profile involving a life expectancy gain of six months is considerably more likely to be chosen than an otherwise identical profile involving a life expectancy gain of three months. The coefficient for the 'alternative' end of life dummy (defined in terms of life expectancy without treatment but not life expectancy gain) was small and not statistically significant. This suggests that any observed support for NICE's end of life policy amongst this sample requires that the policy includes a life extension criterion.

5.5 Conclusions

This chapter has described a large-scale DCE that examined whether the policy of giving higher priority to life-extending end of life treatments (as specified by NICE) than to other types of treatments is consistent with the preferences of members of the general public. The results provide little evidence of support for an end of life premium. When asked to make decisions about the treatment of hypothetical patients with relatively short life expectancies, most respondents' choices were driven by the size of the gains offered by treatment.

Given the aim of the study, the DCE method was useful in that it facilitated the inclusion of multiple levels for each attribute, including levels smaller than, equal to, and larger than those implied by NICE's end of life criteria. The analytical approach also allowed choice probabilities to be predicted for all possible combinations of attributes and levels. However, due to concerns about the complexity of the experimental design and the resulting choice sets, only four attributes were included in the standard DCE tasks. This meant that preferences regarding preparedness had to be examined using separate extension tasks (potentially leading to focusing effects) and other attributes of interest – such as patient age – were omitted altogether. Unlike in empirical studies 1 and 2, no indifference option was available to respondents. On the other hand – and consistent with empirical studies 1 and 2 – all tasks used diagrams to present information to respondents. The next, and fourth, empirical study (Chapter 6)

examines whether people's stated preferences regarding end of life treatments are sensitive to these framing choices and to other study design considerations. Empirical study 4 also further investigates the issue of preparedness, and addresses a key limitation of empirical study 3 by using attitudinal questions to assess whether respondents agree with the policy implications of their responses to trade-off tasks.

6 VALUING HEALTH AT THE END OF LIFE: AN EXAMINATION OF FRAMING EFFECTS AND STUDY DESIGN CONSIDERATIONS (EMPIRICAL STUDY 4)

6.1 Introduction

The literature review (Chapter 2) identified several studies reporting evidence consistent with an end of life premium, and a similar number of studies reporting evidence not consistent with an end of life premium. 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 author's own studies, none of the studies reviewed attempted to control for time-related preferences. The findings of empirical studies 1, 2 and 3 (Chapters 3, 4 and 5, respectively) all suggest 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. The issue of preparedness has generated interest amongst academic and industry audiences during presentations and discussions of the findings of empirical studies 2 and 3 (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 (2013) and McHugh *et al.* (2015), 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[20] and operationalised. For example, studies that included visual

[20] Following Plous (1993), 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 (Luchini and Watson, 2013). 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.

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 (Dolan *et al.*, 2003; Tsuchiya and Watson, forthcoming). 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 findings of the review, coupled with the fact that empirical studies 2 and 3 reported qualitatively different results, provide the motivation for empirical study 4, reported in this chapter. This study seeks to address the same overall research question as the previous studies, whilst additionally testing several hypotheses based on research questions arising from the literature review.

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.

6.2 Methods

6.2.1 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 empirical study 3 (see 5.2.1.5). 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

See Appendix 16 for an outline of the survey with screenshots of each element.

The primary method used in this study was a choice exercise similar to that used in empirical studies 1 and 2. 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 (as was the case for empirical study 3), but it is less suitable when

testing hypotheses regarding the isolated impact of a large number of attributes and study design approaches.

6.2.1.1 Scenarios S1 to S7

Following the approach used in empirical studies 1, 2 and 3, 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 (%)[21]
- 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 6.2.1.6).

Table 6-1 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).

[21] Note that, as with the 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.

Table 6-1. 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 of empirical study 2 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 (Mason *et al.*, 2011). 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 (Rowen *et al.*, 2016a), 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.

6.2.1.2 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 (2013), 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.

6.2.1.3 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

6.2.1.4 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 6.2.1.6). 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' (Cummings and Taylor, 1999) 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 (Özdemir *et al.*, 2009).

6.2.1.5 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.* (2016a; see Rowen *et al.*, 2014 for full details) and Shah *et al.* (2015b), 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 6.2.5).

6.2.1.6 Study design

Respondents were randomly allocated to one of six versions of the survey (Table 6-2). 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 in empirical study 3, which in turn had adapted the design of an existing survey used by Rowen *et al.*, 2016a) 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 6-1 and Figure 6-2 for screenshots showing how the initial question for S1 was presented in the visual aid arm and the no visual aid arm, respectively.

Table 6-2. 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 6-1. Screenshot from survey (S1; visual aid arm; forced choice arm)

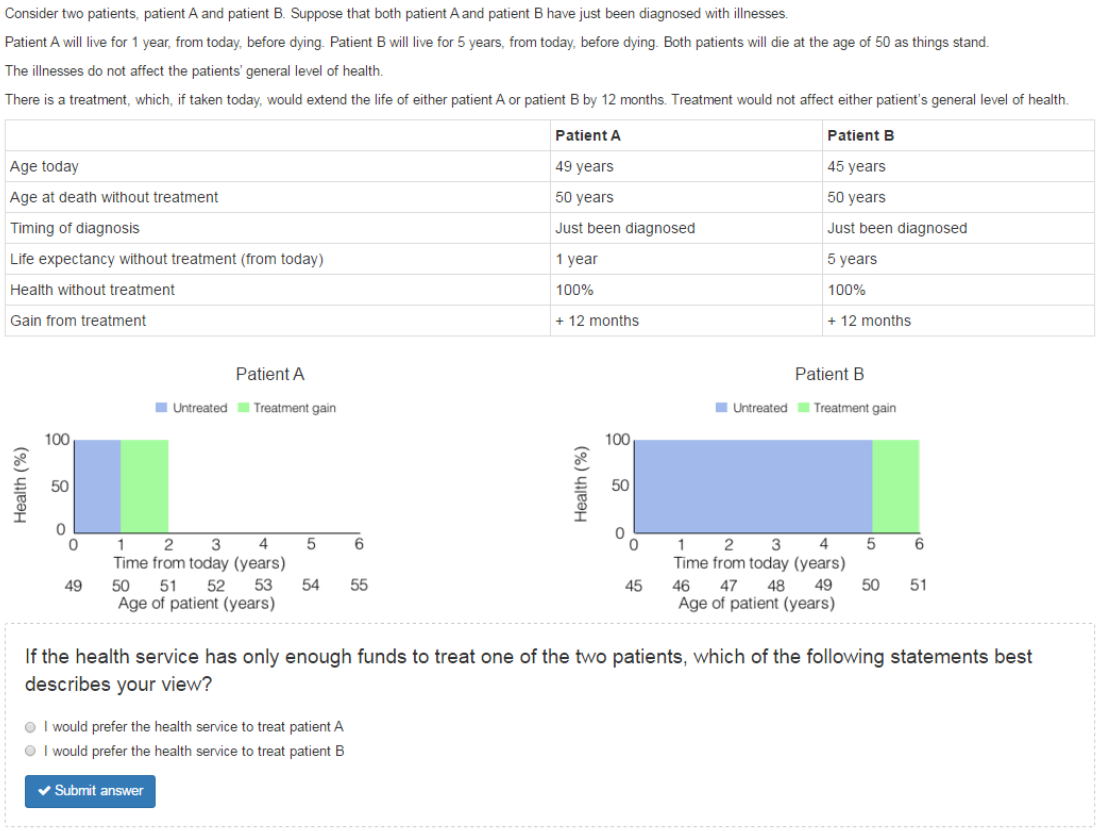
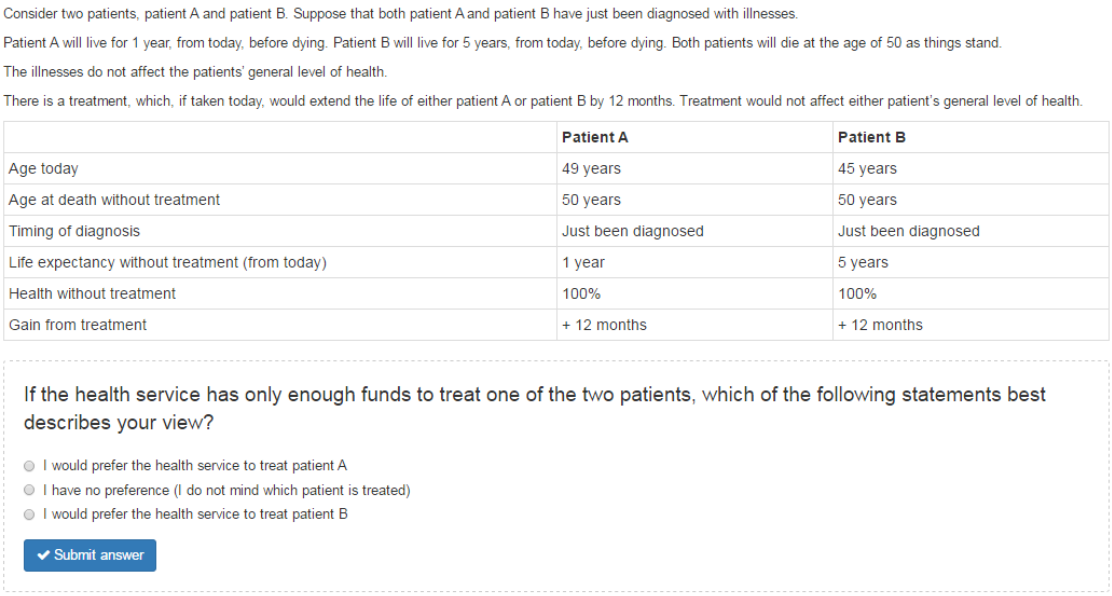


Figure 6-2. Screenshot from survey (S1; no visual aid arm; indifference option 1 arm)



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 6-1 shows the choices available in the forced choice arm, and Figure 6-2 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.

6.2.2 Administration of survey

An internet survey (as used in empirical study 3) was used in favour of face-to-face interviews (as used in empirical studies 1 and 2) 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 in 5.2.2.

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. The same panel was used to recruit respondents for empirical study 3. As with empirical study 3, 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.

6.2.4 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.

6.2.5 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 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 author observing but not assisting or interfering. Following an interview guide (Appendix 18), the author 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.

6.2.5.1 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.

6.2.5.2 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 author 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.

6.2.6 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. This is consistent with the approach used in empirical studies 2 and 3. Analyses were undertaken using the Microsoft Excel and Stata (StataCorp, 2013) software packages.

6.3 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 were 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 6-3). 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 6-3. 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 6-4) (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 6-4. 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 6-5 presents the background characteristics of the sample. The sample was representative of the general population with respect to age, gender and social grade (Office for National Statistics, 2011; National Readership Survey, 2015). The study arms were well balanced in terms of their composition.

Table 6-5. 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	Empirical study 2
		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%	18%
	30-44	633	26.4%	225	27.9%	408	25.6%	323	26.9%	310	25.9%	26%	27%
	45-59	597	24.9%	178	22.1%	419	26.3%	302	25.1%	295	24.6%	25%	22%
	60+	694	28.9%	239	29.6%	455	28.5%	336	28.0%	358	29.9%	28%	33%
Gender	Female	1235	51.4%	429	53.2%	806	50.6%	615	51.2%	620	51.7%	51%	51%
	Male	1166	48.6%	378	46.8%	788	49.4%	587	48.8%	579	48.3%	49%	49%
Social grade [a]	A	93	3.9%	26	3.2%	67	4.2%	42	3.5%	51	4.3%	4%	6%
	B	534	22.2%	176	21.8%	358	22.5%	271	22.5%	263	21.9%	23%	28%
	C1	745	31.0%	260	32.2%	485	30.4%	381	31.7%	364	30.4%	27%	29%
	C2	525	21.9%	174	21.6%	351	22.0%	267	22.2%	258	21.5%	21%	16%
	D	290	12.1%	97	12.0%	193	12.1%	138	11.5%	152	12.7%	16%	9%
	E	214	8.9%	74	9.2%	140	8.8%	103	8.6%	111	9.3%	9%	12%
Household composition	With children	765	31.9%	245	30.4%	520	32.6%	388	32.3%	377	31.4%		24%
	Without children	1636	68.1%	562	69.6%	1074	67.4%	814	67.7%	822	68.6%		76%
Education	None beyond min. school leaving age	559	23.3%	181	22.4%	378	23.7%	286	23.8%	273	22.8%		22%
	Beyond min. school leaving age; no degree	768	32.0%	243	30.1%	525	32.9%	387	32.2%	381	31.8%		31%
	Beyond min. school leaving age; degree	1074	44.7%	383	47.5%	691	43.4%	529	44.0%	545	45.5%		46%
Self-reported general health	Very good	507	21.1%	181	22.4%	326	20.5%	248	20.6%	259	21.6%		25%
	Good	1144	47.6%	377	46.7%	767	48.1%	588	48.9%	556	46.4%		49%
	Fair	575	23.9%	192	23.8%	383	24.0%	291	24.2%	284	23.7%		19%
	Poor	157	6.5%	52	6.4%	105	6.6%	71	5.9%	86	7.2%		5%
	Very poor	18	0.7%	5	0.6%	13	0.8%	4	0.3%	14	1.2%		1%
Experience of terminal illness in friends/family	Yes	1513	63.0%	507	62.8%	1006	63.1%	766	63.7%	747	62.3%		68%
	No	803	33.4%	277	34.3%	526	33.0%	394	32.8%	409	34.1%		30%
	Question skipped	85	3.5%	23	2.9%	62	3.9%	42	3.5%	43	3.6%		2%

[a] Refers to the occupation/qualifications/responsibilities of the chief wage earner of the respondent's household; see National Readership Survey (2015).

6.3.1 Aggregate responses to scenario questions

Table 6-6 to Table 6-13 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 6-6 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 6-6. 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 6-7 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 6-7. 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 6-6 with Table 6-8 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 6-8. 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 6-9 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 6-9. 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 6-9 with Table 6-10 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 6-10. 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 6-11 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 6-11. 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 6-12 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 6-12. 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 6-6 and Table 6-13 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 6-13. 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%)

6.3.2 Impact of experimental modifications

6.3.2.1 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.

6.3.2.2 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.

6.3.3 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

See Appendix 20 for figures summarising responses to the follow-up questions.

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 6-14) and across arms.

Table 6-14. Summary of responses to follow-up questions, by scenario

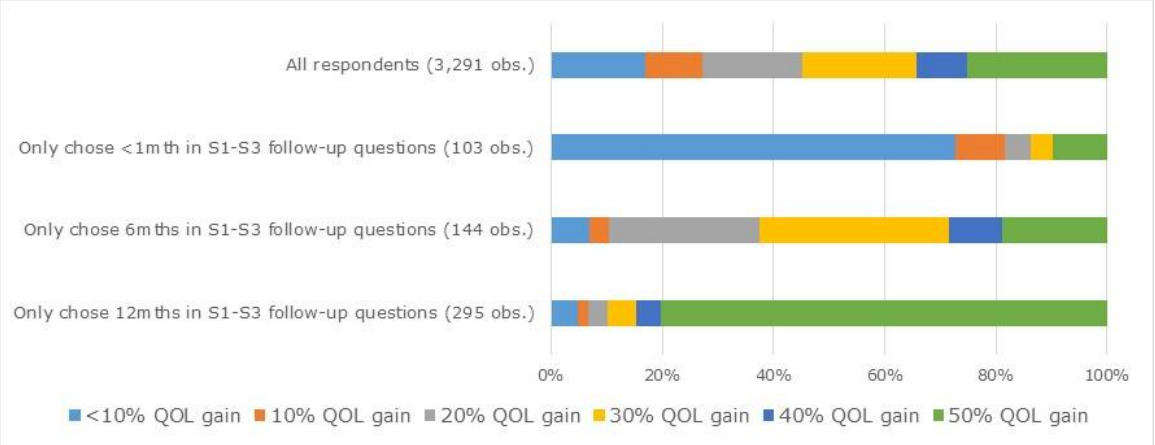
Scenario	Initial choice		Follow-up choice - size of gain		
	Choice	Gain	Mean	Median	Mode
S1	A	12 months	5.9	6.0	6.0
	B	12 months	6.8	6.0	12.0
S2	A	12 months	6.3	6.0	12.0
	B	12 months	6.4	6.0	12.0
S3	A	12 months	6.2	6.0	6.0

Scenario	Initial choice		Follow-up choice - size of gain		
	Choice	Gain	Mean	Median	Mode
	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 6-3 shows 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 6-3. Responses to S4-S6 follow-up questions made by respondents who initially chose the quality of life improvement option in S4-S6



6.3.4 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 6-15, together with possible (face-value) explanations of those choices.

Table 6-15. 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 6-16 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 6-16. 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%)

		III-III-I	III-III-B
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 6.3.7for 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 6.3.6 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.

6.3.5 Cross-tabulations of responses from selected pairs of scenarios

Table 6-17 to Table 6-22 provide cross-tabulations of response data from selected combinations of scenarios that can be used to test some of the hypotheses set out in 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 6-17. 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 6-18. 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 6-19. 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 6-20. 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 6-21. 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 6-22. 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.

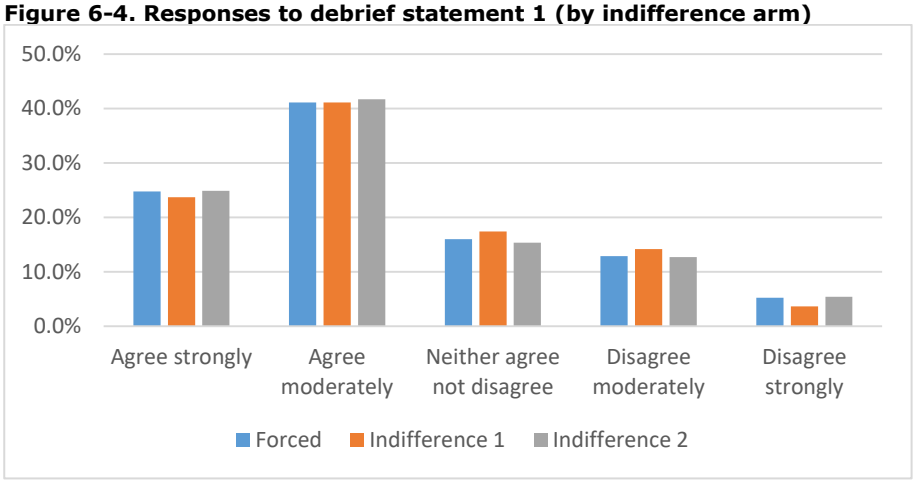
Although a larger proportion of survey version 5 respondents expressed indifference in S8 (165 respondents; 41.6%) than in S1 (158 respondents; 39.8%), the association between perspective and the propensity to express indifference is not statistically significant (chi-squared test; p=0.61). Hence, the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

6.3.6 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).

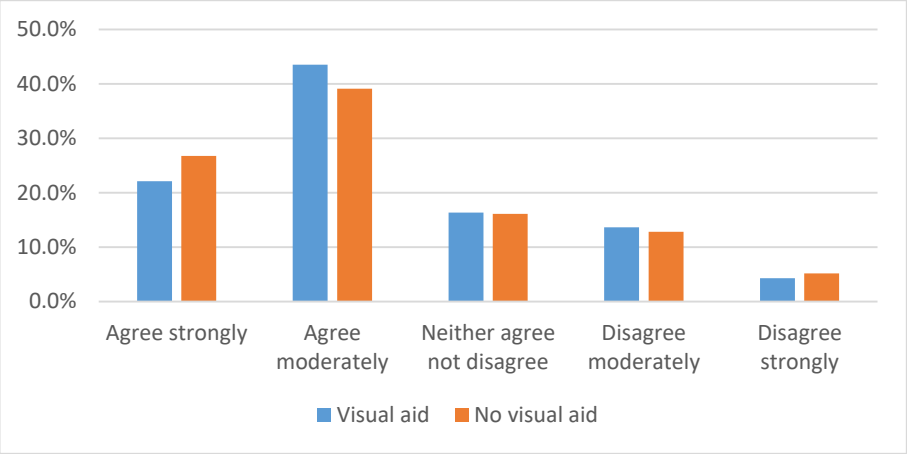
The extent to which respondents agreed or disagreed with debrief statement 1 did not vary greatly by study arm (Figure 6-4 and Figure 6-5). On the other hand, respondents in the forced choice arm were more likely to agree with debrief statement 2 than those who were given an indifference option (Figure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

Figure 6-4. Responses to debrief statement 1 (by indifference arm)



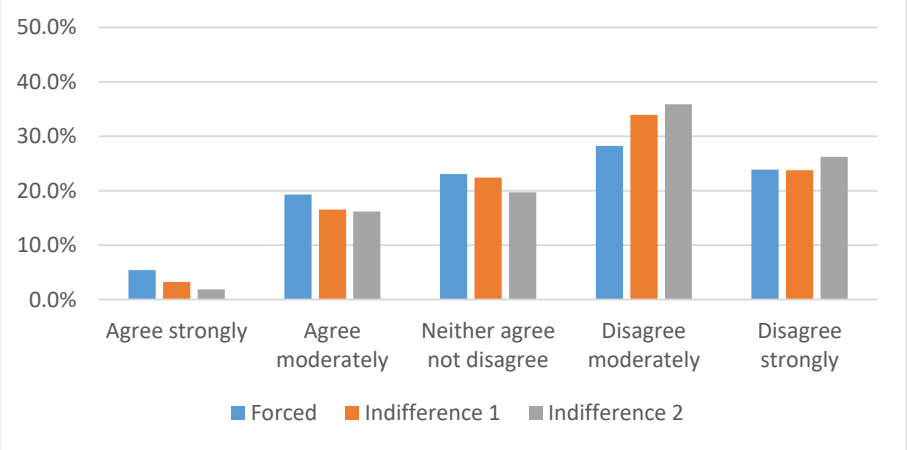
Statement 1: I found it difficult to decide on my answers to the questions

Figure 6-5. Responses to debrief statement 1 (by visual aid arm)



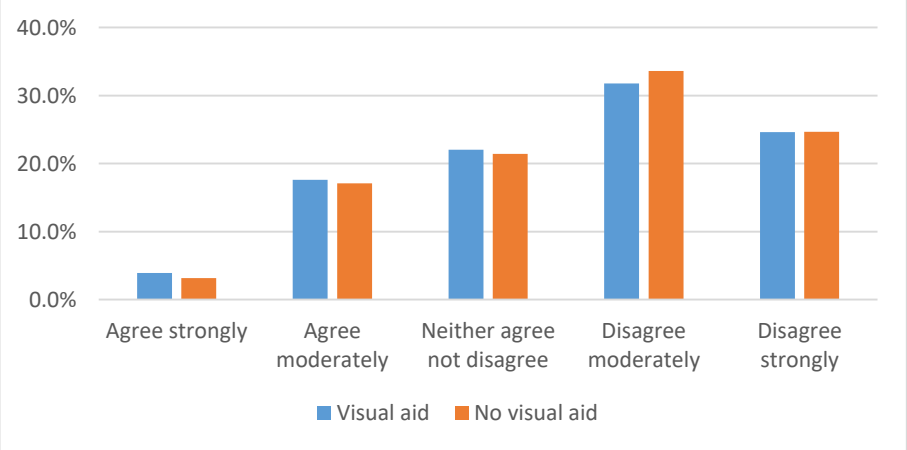
Statement 1: I found it difficult to decide on my answers to the questions

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



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

Figure 6-7. 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).

6.3.7 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 6-23). Excluding these respondents would therefore *strengthen* the finding that giving priority to the treatment of end of life patients is not supported (Table 6-24).

Table 6-23. 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 6-24. 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%

6.3.8 Attitudinal questions

Responses to the six attitudinal questions are shown in Table 6-25. 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 6-25. 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 6-26 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 6-26. Cross-tabulation: S1 vs. attitudinal statement 1

	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 6-27,

Table 6-28 and Table 6-29 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 6-27. 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 6-28. 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 6-29. 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%)
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.

6.3.9 Impact of respondent background characteristics in S1

The results of the multiple linear regression are shown in Table 6-30. 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 6-30. 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

6.3.10 Ordering effects

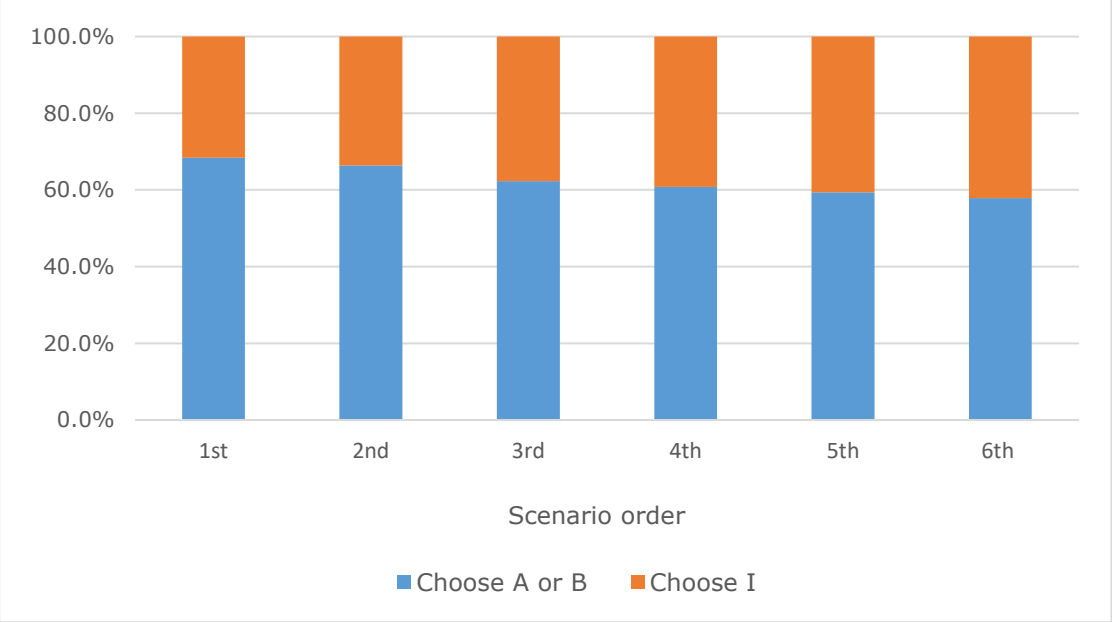
The order in which scenarios S1 to S6 were presented was randomised for each respondent. Table 6-31 shows how the responses made differed depending on whether or not the scenario in question was the first to be presented. In each column, the modal choice is emboldened. In S1 and S3, the modal choice 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 6-31. 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 6-8).

Figure 6-8. Proportion choosing A or B versus proportion choosing I, by scenario order



6.4 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 (Dolan and Shaw, 2004; Stahl *et al.*, 2008; Richardson *et al.*, 2012). NICE's general principle is that health care resources cannot be allocated or restricted on the basis of age (NICE, 2008c; Shah *et al.*, 2013a; Rowen *et al.*, 2016b). 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 (1998) 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 literature review (see 2.4.1). 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.* (2013)). 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 (see 2.4.3) 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, empirical studies 1, 2 and 3 in this thesis; Pennington *et al.*, 2015; Rowen *et al.*, 2016a; Rowen *et al.*, 2016b), 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 (1999) 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' (Green, 2009), 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 (2013), 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 (2013), 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 (Ternent and Tsuchiya, 2013; Oppe *et al.*, 2016).

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.* (2014) 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 (Messick, 1967) – 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, 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 (Sheldon, 1999).

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 in 5.2.2.

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 (see 2.4.5). 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.

6.5 Conclusions

This chapter 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.

The ways in which the results of this study relate to those of the other empirical studies are discussed in Chapter 7.

7 DISCUSSION

7.1 Introduction

This chapter discusses the findings of the overall thesis. It begins by summarising the main contributions of the research. It then highlights some of the ways in which the empirical studies complement and differ from both each other and other published research on the topic. The limitations and ambiguities of the research are then discussed. The chapter concludes by presenting recommendations for further research.

The thesis was motivated by NICE's introduction of supplementary guidance for the appraisal of life-extending end of life treatments (see 1.8.2), and by calls for research on the extent of societal support for such a policy (see 1.9). It set out to address the following research question: *do 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?* A series of empirical studies were used to try to answer this question, focusing on the preferences of the UK general public and the definition of end of life adopted by NICE in its supplementary guidance.

7.2 Key contributions

The key contributions of the research are loosely organised into three (not necessarily mutually exclusive) categories: (1) contributions in terms of topics examined; (2) contributions in terms of methodological approaches; and (3) contributions in terms of important findings.

7.2.1 Topics examined

7.2.1.1 Specific focus on NICE's end of life policy

The empirical studies reported in this thesis were amongst the first to have directly addressed research questions arising from NICE's policy on the appraisal of life-extending end of life treatments. All four studies presented choices between treatments that *would* meet NICE's criteria for special consideration (life expectancy without treatment of less than 24 months and life extension of greater than three months) and treatments that *would not*. Empirical study 3 went further by including in the regression analysis a dummy variable defined in terms of NICE's cut-offs and by analysing the impacts of alternative cut-offs.

All of the empirical studies also examined whether life-extending end of life treatments are preferred to quality of life-improving treatments. Given that the NICE's end of life criteria accommodate life extensions but not quality of life

improvements *per se*,[22] this seems like a relevant research question. Yet the literature review showed that evidence on this particular issue is scarce (see 2.3.8.4).

7.2.1.2 Examination of preparedness

The literature review revealed that the issue of preparedness has not been examined in most of the studies published to date.[23] The research in this thesis has therefore made a novel contribution to the understanding of end of life-related preferences in this respect. Further research to validate or extend the findings regarding preparedness in this thesis would be welcomed.

7.2.1.3 Novelty of the literature review

The literature review is novel in that no comparable reviews of public end of life-related preferences are available. Existing general reviews of the empirical ethics literature that had sought to include end of life as an attribute of interest had reported finding no or limited relevant studies (Green, 2011; Keetharuth *et al.*, 2011). Three of the older studies identified in the end of life review (Dolan and Cookson, 2000; Dolan and Tsuchiya, 2005; Stolk *et al.*, 2005) had also been included in the author's earlier review of public preferences regarding severity of illness as a priority-setting criterion (Shah, 2009). However, most of the studies in the severity review described severity in terms of quality of life rather than life expectancy; and most of the studies in the end of life review were conducted and published after NICE issued its supplementary guidance in 2009. Given the increasing interest in the relative value of life extensions for patients with short life expectancy, the need for an up-to-date and specific review was clear.

7.2.2 Methodological approaches

7.2.2.1 Examination of preparedness

A novel attempt was made in empirical studies 1 and 2 to design a scenario involving a choice between treating an end of life patient and treating a non-end of life patient whilst controlling for time-related preferences. This involved describing one patient as having been diagnosed with their illness nine years prior to the start

[22] It is worth noting that the NICE end of life policy asks appraisal committees to consider assuming that the life extensions generated by eligible treatments are experienced in full quality of life (Box 1). This aspect of the guidance was not examined in this thesis as it was judged that it simply described one possible mechanism for upward-adjusting QALY gains, and was not intended to reflect societal preferences.

[23] An exception is McHugh *et al.* (2015), in which a statement referring to the notion ('It is important to give a dying person and their family time to prepare for their death, put their affairs in order, make peace and say goodbyes') was included in the statement set. Respondents in that study identifying with the shared perspective emphasising patient choice and the right to life-extending treatment were likely to agree with this statement.

of the scenario and the other patient as having just been diagnosed. Life expectancy without treatment (from the start of the scenario) was the same for both patients. An observed preference for treating the latter patient could be explained by concern about how little time that patient will have had to prepare for death.

Empirical studies 3 and 4 also investigated the issue of preparedness by presenting respondents with scenarios in which one of the patients had known about their illness for some time (two years and five years, respectively) and the other patient had only just learned of their illness. In many cases, the patient with longer life expectancy without treatment (from the start of the scenario) would have known about their prognosis for less time than the patient with shorter life expectancy. This meant the respondents in these studies were being asked to make trade-offs between prioritising based on concerns about end of life and prioritising based on concerns about preparedness.

7.2.2.2 Examination of indifference

An important methodological consideration in the design of stated preference studies is whether or not to permit respondents to express indifference between the available options. The literature review (Chapter 2) showed a fairly even split between the number of studies in which indifference could be expressed and number in which it could not. Yet the use or otherwise of indifference options was rarely discussed or justified in any of the articles.

How indifference options are framed also matters. For example, framed one way, an indifference option may appeal to respondents who consider all of the alternatives on offer to be attractive (and equally so); framed another way, it may appeal to respondents who do not wish to engage in the choice task. Elsewhere in the empirical ethics literature, Ubel (1999) compared forced (pairwise) choice priority-setting questions with (otherwise identical) questions in which a third option of dividing resources equally was available. It was found that the majority of respondents expressed indifference when given the opportunity to do so. Building on Ubel's study design, and drawing on another study by Oddsson (2003), Green (2009) tested the addition of a fourth option in which respondents could indicate that they were unable to make a decision and would prefer that the choice be made by others. Green reported that very few respondents chose this fourth option, and concluded that when respondents state a preference for equality it may be interpreted as a true preference rather than a means of avoiding making difficult decisions.

The research in this thesis adds to the contributions of Ubel and Green. The empirical studies analyse and discuss the impact of including or omitting an indifference option, and of alternative framings of indifference options.

7.2.2.3 Examination of the impact of study perspective

The majority of studies included in the literature review asked respondents to adopt a social decision-maker perspective. Four studies used an individual perspective in which respondents were explicitly asked to imagine that the hypothetical choices applied to their own lives. Two of those four studies (Shiroiwa *et al.*, 2010; Pinto-Prades *et al.*, 2014) compared both types of perspective by asking respondents to complete individual perspective tasks and corresponding (but separate) social decision-maker perspective tasks. All of the studies that sought individual perspective preferences did so by applying the willingness-to-pay technique – a method that is rarely applied in social decision-maker perspective studies. Hence, it is difficult to assess whether any observed differences in results between the two types of task were driven by differences in study perspective *per se* or by differences in the 'payment vehicle' (i.e. what was being traded – money or some other benefit).

The empirical studies reported in this thesis were largely undertaken from a social decision-maker perspective. In empirical study 4, however, an attempt was made to compare end of life-related preferences elicited using both types of perspective through the inclusion of scenario S8. The approach used was novel in that it used a method other than willingness-to-pay to examine individual perspective preferences regarding end of life treatments. It may be that by presenting two tasks that differed in terms of perspective but were otherwise intended to be identical, and by avoiding explicit consideration of money from the tasks, empirical study 4 managed both to overcome some of the limitations of willingness-to-pay studies in determining decisions about the allocation of public resources (see 2.4.1), and to isolate the effect of the change in perspective from the effect of the change in payment vehicle.

The author is aware of two recent, unpublished studies that have also attempted to use methods other than willingness-to-pay to examine individual perspective preferences for an end of life premium. Gyrd-Hansen *et al.* (2017) asked respondents to choose which they would prefer out of four free extensions of a hypothetical private insurance policy. Nielsen *et al.* (2017) applied a risk-risk trade-off approach which involved asking respondents to choose between a reduction in mortality risk and an improvement (both a life extension and a quality of life improvement) to their end of life situation.

7.2.2.4 Searching for search terms in the literature review

The literature review applied a creative approach to searching for search terms and a comprehensive approach to extracting data that were of relevance to the research question. At the time of conduct, the number of studies that specifically set out to examine end of life-related preferences remained limited – only eight of the identified articles explicitly mentioned end of life, or some synonym for end of life, in the stated study objectives. A contribution of the review is that the author extended the scope in order to capture studies that set out to answer an altogether different research question but happened to analyse or report data that enabled the comparison of preferences regarding the treatment of (or of values of gains for) patients with different life expectancies (Stolk *et al.*, 2005; Richardson *et al.*, 2012; Rowen *et al.*, 2016b).

7.2.3 Important findings

7.2.3.1 Choices running counter to an end of life premium

The results varied from study to study, but overall the evidence presented in this thesis suggests that people do *not* place greater weight on a unit of health gain for end of life patients (as defined by NICE) than on that for other types of patients. An important finding in all four empirical studies was that non-trivial proportions of respondents favoured the treatment of the non-end of life patient over the end of life patient, in many cases even when an indifference option was available. In empirical study 1, three of the 21 respondents (14.3%) chose to treat the non-end of life patient in scenario S1. Comments made by these respondents when answering the probing questions suggest that they felt that the life extension should be given to someone who has a better opportunity to make the most out of the additional time offered by treatment. In empirical study 2, 13 of the 50 respondents (26.0%) made the same choice, and their responses to the follow-up tick-box tasks suggest that this was driven by a belief that patients with longer life expectancy are better placed to make the most out of a short life extension. In empirical study 3, the patient with longer life expectancy was given priority over the patient with shorter life expectancy 55% of the time across all choice sets. Three-hundred and fifty-five of the 3,969 respondents (8.7%) *never* chose to give priority to the patient with shorter life expectancy. In empirical study 4, 803 of the 2,401 respondents (33.4%) chose to treat the non-end of life patient in scenario S1.

Such results had not been anticipated at the start of the PhD research programme. Placing greater weight to a unit of health gain for patients with longer life

expectancy than on that for patients with shorter life expectancy, *ceteris paribus*, is inconsistent not only with NICE's end of life policy, but also with the QALY-maximisation framework and many other candidate approaches for determining how health care resources should be allocated. The thesis includes some attempts to explain these preferences, drawing on respondents' answers to follow-up questions (see 3.4.1 and 4.3.1) and on findings reported elsewhere that respondents may be acting strategically, overcompensating to counteract the choices they anticipate that others will make (Shah *et al.*, 2015b). The potential for alternative research methods to better understand such 'unexpected' preferences is discussed in 7.5.1.1.

7.2.3.2 Findings regarding the use of indifference options

In empirical study 2, the indifference option (indicating that the respondent had no preference over which patient should be treated) was presented more explicitly and with greater visual prominence than in empirical study 1. Indifference was expressed more frequently overall, and by a larger proportion of respondents, in empirical study 2 than in empirical study 1. Empirical study 4 provides evidence that the findings of a survey of end of life-related preferences may depend on whether and how indifference can be expressed: in scenario S1, for example, the modal choice in the forced choice arm was to treat the end of life patient, whereas for respondents in the indifference arm this was the *least* common choice. If an end of life policy were to be made based on the findings of empirical study 4 (taken at face value) and on majoritarian grounds, then the decision about whether to include two or three response options could be a determining factor.

On a related note, the responses to the follow-up questions in empirical study 4 suggest that when faced with a large number of response options, respondents tend to restrict their choices to a small subset of those options, such as the middle option. This could explain why the indifference option in the study by Linley and Hughes (2013) was selected so frequently. In that study, 11 response options were available but the single option implying indifference – equal allocation to both patient groups – accounted for nearly half of all choices made (in some of the other scenarios addressing factors other than end of life, the proportion of respondents expressing indifference was even greater). In reporting their results, Linley and Hughes collapsed the 11 response options into three categories. Recommendations for further investigation of indifference expression in stated preference research are provided in 7.5.1.4.

7.2.3.3 Findings regarding preparedness

Responses to the probing questions (in empirical study 1) and the tick-box tasks (in empirical study 2) suggest that the amount of time patients have to prepare for death was a consideration for a number of respondents. The results from both studies suggest that any observed preference for treating an end of life patient may be driven by concern about how long they have known their prognosis rather than by concern about how long they have left to live.

When asked to choose between a patient who had known about their illness for some time and another patient who had only just learned of their illness, empirical studies 3 and 4 both reported an increase in the proportion of respondents choosing to treat the latter patient compared to when no 'time with knowledge' information was provided.

7.2.3.4 Quality of life-improving versus life-extending end of life treatments

Empirical studies 1, 2 and 4 report findings to suggest that quality of life-improving treatments are more highly valued than life-extending treatments for end of life patients. This is consistent with the findings of Pinto-Prades *et al.* (2014), the only study identified in the literature other than the author's own studies to have reported findings relating directly to this issue. To the author's knowledge, empirical study 3 is the only study to have reported evidence of public support for prioritising life-extending end of life treatments over quality of life-improving end of life treatments in a resource allocation context.

7.2.3.5 State of the literature

The key finding of the literature review (which included publications based on empirical studies 2 and 3 of this thesis) was that the evidence is mixed, with similar numbers of studies reporting evidence consistent with and not consistent with an end of life premium. There are a number of reasons why studies may differ in terms of their findings: for example, different samples (often reflecting different populations), different study objectives, and the use of different methodologies. Drawing on the results of the review, some hypotheses based on the notion that preferences regarding an end of life premium may depend on study design considerations, such as study perspective and the use of indifference options and visual aids, were tested in empirical study 4. However, it is also notable that several authors reported evidence of heterogeneous preferences within their own individual studies, controlling for sampling frame, study objective and methodology (for example, Pinto-Prades *et al.*, 2014; McHugh *et al.*, 2015; Shah *et al.*, 2015).

This lends weight to a claim made in a recent NICE Citizens' Council report that there are no straightforward answers to questions about societal values because of the diversity of the population and range of opinions within society (NICE, 2014b). A pertinent question, then (and not one that this thesis sought to answer), is how the findings of a given study should be reported in order to convey relevant information about the preferences of the sample. Is it sufficient simply to report a single representative preference (such as that of the median respondent), or is it important to account for the heterogeneity of views expressed? Devlin *et al.* (2017) suggest that there are no strong normative grounds for any single measure of overall preference in social choices.

7.3 Differences between the studies in this thesis

The four empirical studies reported in this thesis were all designed by the same author, with the same overarching research question in mind. It is therefore unsurprising that a number of features and characteristics are common across all four studies. However, each study had its own unique focus, and the design of the later studies was informed by the findings of and challenges encountered in the earlier studies. This section identifies and discusses a selection of key differences between the studies. Full details are available in Appendix 21, which summarises the four studies in terms of the same data extraction fields as used in the literature review (Table 2-1).

7.3.1 Distinction between end of life and non-end of life patients

Respondents in empirical studies 1 and 2 were more likely to choose to treat the end of life patient than were respondents in empirical studies 3 and 4. Empirical studies 1, 2 and 4 all included scenarios that sought to present a straightforward comparison between an end of life patient and a non-end of life patient. While the prognosis of the end of life patient was the same in all three studies, the prognosis of the non-end of life patient varied. In empirical studies 1 and 2, the non-end of life patient had a life expectancy of 10 years without treatment. By comparison, the non-end of life patient in empirical study 4 had a life expectancy of just five years. Similarly, the longest level used for the life expectancy without treatment attribute in empirical study 3 was also five years. Both choices of level are consistent with the designs of other studies in the literature (Shiroiwa *et al.*, 2010; Abel Olsen, 2013; Linley and Hughes, 2013; Skedgel *et al.*, 2015). While five years of life expectancy is well beyond the two-year cut-off in the NICE criteria, it is nevertheless a poor prognosis and may well be considered by many respondents to

represent an end of life situation. Hence, the difference between the end of life patient and the non-end of life patient was starker in empirical studies 1 and 2. This could explain why the results of the earlier studies differed from those of the later studies.[24]

While a life expectancy of 10 years is less likely to be considered an end of life situation than a life expectancy of five years, it has been argued that *any* presentation of a known life expectancy – no matter how long – may be interpreted as a 'life sentence' and may therefore induce end of life-type preferences (Gyrd-Hansen *et al.*, 2017). Studies using alternative comparators such as scenarios involving temporary quality of life losses (Shiroiwa *et al.*, 2013; Pinto-Prades *et al.*, 2014; Pennington *et al.*, 2015) are informative because the end of life scenario is compared to scenarios that do not involve a premature death. This reflects the broad range of alternative ways in which the resources required to provide end of life treatments could otherwise be used. Similarly, gains generated by end of life treatments could be compared to gains achieved through the prevention of illness, or through reductions in lifetime mortality risk. Such comparators have been tested in two recent, unpublished studies (Gyrd-Hansen *et al.*, 2017; Nielsen *et al.*, 2017).

7.3.2 Influence of patient age

A statistically significant association between patient age and the propensity to prioritise the treatment of the end of life patient was observed in empirical study 4. By comparison, empirical study 2 did not find that respondents' choices were influenced by the ages of the patients. This is likely due to the fact that in empirical study 4, the difference in the ages of the two patients in the scenario of interest was much larger (20 years; the patients would die aged 70 years and 50 years without treatment, respectively) than the corresponding difference in empirical study 2 (nine years; absolute ages were not specified).

The finding in empirical study 1 that respondents had made different assumptions about the ages of the patients informed subsequent decisions both to specify that the patients were both adults in empirical studies 2 and 3, and to specify the exact ages of the patients in empirical study 4. Researchers should consider providing information about patient ages when developing hypothetical priority-setting scenarios. This would reduce the likelihood of respondents making inappropriate or unintended assumptions, such as imagining that the patients are children when the researcher is interested in eliciting preferences regarding the treatment of adults.

[24] It should be noted, however, that Abel Olsen (2013) and Skedgel *et al.* (2015) also presented a maximum life expectancy without treatment of 10 years, and neither reported evidence consistent with an end of life premium

7.3.3 Support for QALY-maximisation

In empirical studies 1, 2 and 4, very few respondents made sets of choices that corresponded perfectly to those of a strict QALY-maximiser. By comparison, the modelling results in empirical study 3 demonstrate that QALY gains were the primary driver of respondents' choices in that study, and a sizeable proportion (albeit still a minority) of the respondents in that study consistently followed a strategy of choosing to treat the patient who stood to gain more QALYs from treatment.

This difference in results may be linked to how the size of QALY gain was controlled for. In empirical studies 1, 2 and 4, the scenarios did not allow a trade-off to be made between giving priority to the end of life patient and pursuing a QALY-maximisation approach, since in almost all cases the size of the (undiscounted) QALY gain was held equal for both patients. In principle, a respondent who was *only* interested in maximising QALYs should have expressed indifference in all choices in which both alternatives offered equal-sized QALY gains. In practice, however, respondents may be disinclined to choose an indifference or 'no preference' option, particularly if that option is not presented as prominently as the other options (as was the case in empirical study 1). Further, respondents may deem QALY-maximisation to be an important objective but would still be willing to consider other factors, such as the extent to which the patients are at the end of life, in cases where there is little or no difference in QALY gains between the alternatives. In empirical study 3, on the other hand, the QALY gains differed between patients in the vast majority of the choice sets presented. This allowed direct trade-offs between QALY-maximisation and end of life prioritisation (as well as other objectives and choice strategies) to be made.

7.3.4 Propensity to express indifference

In empirical study 1, indifference was expressed infrequently, accounting for only 13% of all choices made. The findings of empirical study 1 informed the design of empirical study 2, in which the indifference option was presented more explicitly and with greater visual prominence. In that study, indifference was expressed more often, accounting for 20% of all choices. In empirical study 4, indifference was expressed 39% of the time by respondents in study arms in which an explicit indifference option was available, and 26% of the time by respondents in the forced choice arm in which respondents could only express indifference indirectly via their response to the follow-up question. The increased tendency to express indifference in empirical study 4 might reflect the fact that the difference between the

alternatives was (in some scenarios) less pronounced than was the case in the earlier studies (see 7.3.1).

These findings indicate that the way in which indifference options are worded and presented matters. It may be the case that respondents are more inclined to express indifference in internet surveys (as used in empirical study 4) than in interviewer-led modes of administration (as used in empirical studies 1 and 2), possibly as a default choice that allows them to avoid taking time to make difficult decision. Indeed, this was one of the rationales for adopting a forced choice design in empirical study 3.

7.3.5 Quality of life-improving versus life-extending treatments

Empirical studies 1, 2 and 4 all reported evidence to suggest that quality of life-improving treatments are preferred to life-extending treatments for end of life patients (with one year of life expectancy without treatment in 50% quality of life without treatment). In empirical study 2 it was shown that this result was unaffected by whether the end of life patients were younger adults or older adults. In empirical studies 2 and 4 it was shown that the preference for quality of life-improving treatments over life-extending treatments persisted (albeit to differing degrees) even when the quality of life-improving treatment would be provided to a patient with longer life expectancy.

The comparison of preferences for quality of life-improving and life-extending treatments was less straightforward in empirical study 3 due to differences in the study design and methodology. Nevertheless, in that study both the regression results and the predicted probability of choice analysis indicate the opposite result – that is, respondents' choices were guided by life extensions to a greater degree than by quality of life improvements. The experimental design for empirical study 3 did not include any choice sets that compared a quality of life-improving treatment with a life-extending treatment whilst controlling for all other aspects (including the size of QALY gain and the extent to which the two patients were at the end of life). The predicted probability analysis suggests that if a choice set had been included in empirical study 3 that corresponded directly to the relevant scenarios in empirical studies 1 (S4), 2 (S4/S5) and 4 (S4), then the patient who stood to gain a life extension would have been chosen more often. However, this is the result of econometric modelling and the various assumptions underpinning the methods of analysis. It is unclear whether a similar result would have arisen if respondents in

empirical study 3 had faced this choice set explicitly, thereby allowing the direct observation of their responses.

7.3.6 Bias towards particular response options

In empirical study 3, it was found that respondents were more likely overall to choose the alternative appearing at the bottom of the screen (labelled patient B). In this study, the labelling of the alternatives was randomised in order to control for such bias due to the positioning of options. Randomisation of labels was not applied in the other studies. Since the visual presentation of choice tasks was very similar in empirical studies 3 and 4 (see Figure 5-1 and Figure 6-1), it is plausible that a similar bias towards patient B would have been present in empirical study 4. This would have had the effect of, *inter alia*, indicating greater support for the non-end of life patient in scenario S1 and for the life-extending treatment in scenario S4 than if the labelling of alternatives had been randomised.

7.4 Comparing the research in this thesis with the research of others

This section highlights some of the ways in which the research in this thesis compares to studies undertaken by other investigators, focusing on three studies of the preferences of the UK general public that were motivated by recent policy developments.

7.4.1 Comparison with the Rowen *et al.* studies

Two studies that shared similar designs and approaches, yet reached different conclusions about the level of support for an end of life premium, were Rowen *et al.* (2016a) and empirical study 3 in this thesis. Both studies used web-based DCEs to examine the views of large samples of the UK population (recruited from panels maintained by market research agencies). Both studies also used forced choice designs and similar visual presentations. Yet empirical study 3 reported regression results that indicated little support for an end of life premium and prioritising based on life expectancy without treatment, while Rowen *et al.* reported robust and consistent support for an end of life premium.

Some differences between the two studies should be mentioned. Whereas empirical study 3 used only one survey version and made no reference to age (either directly or indirectly, other than the instruction that the patients were both adults of the same age), Rowen *et al.* used four different survey versions, each depicting a different 'life expectancy without the condition' (which may have been interpreted by respondents as a proxy for the patients' life stage). The coefficients of the

explanatory variables differed across survey versions, though the authors' main conclusions were based on the pooled data. Both studies used conditional logit regressions to model the choice data, though there were differences in the model specification, reflecting the differing objectives of the studies. For example, in models that included an end of life dummy variable, Rowen *et al.* did not include an explanatory variable representing life expectancy without treatment, based on concerns about conceptual overlap in these variables. However, the differences in conclusions between the two studies cannot be explained by these factors alone, since Rowen *et al.*'s end of life dummy variable was shown to be important in all survey versions, and the lack of importance of life expectancy without treatment in empirical study 3 was demonstrated not only in the regression analysis but also in the descriptive analysis of the raw choice data. Other features of the Rowen *et al.* study that differed from empirical study 3 included: the use of practice questions (intended to introduce respondents to the concept of higher and lower burden of illness); the presentation of choices between patient groups (rather than between individual patients); and the focus on marginal rates of substitution rather than on predicted probabilities in the analysis.

Two other studies that shared similar characteristics were Rowen *et al.* (2016b) and empirical study 4. In each of the studies, a single survey was tested using multiple framings and formats. Rowen *et al.* (2016b) tested two modes of administration (face-to-face interview versus unsupervised internet survey), two sets of question wording (for example, labelling the alternatives in terms of conditions or patient groups), and the use of visual aids and instructional videos. The authors reported that the mode of administration influenced respondents' choices, whereas the other factors did not. In empirical study 4, the impact of study perspective and the use of indifference options and visual aids was examined. All three factors were found to influence responses to some extent, though the primary conclusion (of limited support for an end of life premium) was common to all formats and framings.

It is difficult to conclude from empirical study 4 which approach is most suitable for eliciting preferences regarding end of life prioritisation (and regarding equity and social values more generally). Rather, the findings suggest that it is important to use multiple approaches in order to ensure that the results of a given study are suitably robust and not an artefact of the methodology. This might be considered a rather unsatisfactory recommendation, as the use of multiple survey versions can make a study unwieldy and challenging to manage, and the findings more difficult to communicate. Further, it increases the likelihood that the study will produce results that are conflicting or inconclusive, which makes them less likely to be used to inform policy decisions directly. However, the author's view is that the stated

preference researcher's primary responsibility is not to generate ground-breaking and policy-influencing results, but to contribute to a shared understanding of how best to elicit, analyse and interpret preference data, and to be honest and explicit about the caveats and limitations of their work.

It should be noted that the Rowen *et al.* (2016b) study acted, in effect, as a preliminary study that informed the design of the larger-scale Rowen *et al.* (2016a) study. This is a potentially useful approach: pilot several candidate versions of the survey in a preliminary study, and use the results to inform which version(s) to use in the main study. If two different methods or formats generate similar choice data, this gives the researcher freedom to select a preferred approach based on factors such as cost, parsimony or respondent feedback. A more general point is that there are many different, and legitimate, ways of testing a given hypothesis or research question. Given that the results of empirical study 4 differed depending on which specific framing was used, and differed from those of empirical study 2 (which was similar in many respects), it is unsurprising that different conclusions are reached by studies adopting vastly different methodologies and – in many cases – seeking to address very different research questions.

A feature shared by empirical studies 3 and 4 and the two Rowen *et al.* studies was the inclusion of choices in which both of the patients had the same amount of life expectancy and quality of life without treatment, and one patient would gain more from treatment than the other. Under the assumption that health improvements result in positive (or at least non-negative) utility gains, these choice sets may be described as comprising a dominant and a dominated option. Although Lancsar and Louviere (2006) recommend that researchers refrain from excluding data that do not conform to their own preferences and expectations, it is common in health state valuation research to apply exclusion criteria based on so-called 'logical inconsistencies' (Engel *et al.*, 2016). In empirical studies 3 and 4, a minority of respondents (8% and 8%, respectively) chose the dominated option. Sensitivity analyses showed that excluding these respondents did not affect the overall conclusions of either study. These results are very similar to those of the Rowen *et al.* studies, in which 7% (Rowen *et al.*, 2016a) and 9% (Rowen *et al.*, 2016b) of respondents chose the dominated option.

One of the reasons for including tasks with dominated options in stated preference studies is to identify whether respondents are displaying appropriate levels of engagement and understanding – a particular concern for internet surveys and other non-interviewer-assisted modes of administration. Yet Rowen *et al.* (2016b) did not observe statistically significant differences in responses to the relevant

question across different modes of administration, suggesting that interviewer-assisted studies may be as susceptible to this issue as self-complete internet surveys.

7.4.2 Comparison with the Linley and Hughes study

The results of empirical study 4 can also be compared to those of Linley and Hughes (2013), who similarly used an internet survey to seek the views of a large sample of the UK general public. The question relevant to end of life in the Linley and Hughes survey involved a choice between giving a six-month life extension to patients with 18 months of life expectancy without treatment and giving a six-month life extension to patients with 60 months (five years) of life expectancy without treatment. The basic result was the same in both studies: the most common choice was to express indifference between the options.

By comparison, Linley and Hughes reported that 47.6% of respondents expressed indifference, which is higher than the proportion expressing indifference in any of the empirical study 4 arms for the corresponding question (scenario S1). This may be explained by the framing of the choice task and indifference option. Whereas in empirical study 4 the choice was between two individual patients and the indifference option (where available) implied a lack of preference or willingness to prioritise randomly, Linley and Hughes used a budget allocation approach (where the choice was between two patient *groups*) in which 11 different splits were available, including one that involved an equal allocation to both groups. It has been shown elsewhere that options implying a 50:50 split of resources may be deemed more attractive to respondents than an 'I have no preference' option (Shah and Devlin, 2012) or an option implying an unwillingness to choose (Green, 2009). Recommendations for further exploration of indifference options are discussed in 7.5.1.4.

7.5 Limitations and recommendations for further research

The research reported in this thesis is subject to a number of limitations. Many of these have been identified and discussed in the relevant chapters. This section reflects on some of the limitations of the overall programme of research. The purpose here is to understand how such limitations could be overcome in future research and to explore how the author would proceed (in light of what has been learned in this programme of work) if given the opportunity to answer the research question again (in some cases with greater monetary, intellectual and time resources).

It should be noted that all stated preference research methods, including preference elicitation techniques, modes of administration, design choices and analytical approaches, suffer from limitations and are open to critique to some extent. This PhD research was not intended to investigate all possible methods or to contain an extensive methodological review, though the use of different methods within and between studies has generated insights into the advantages and disadvantages of the various approaches. Drawing on these insights, this section includes conjectures and judgements about what might have been achieved using different research methods.

7.5.1 Methodological approaches

7.5.1.1 Understanding respondents' choices

All four empirical studies reported that a non-trivial proportion of respondents made choices that implied that they placed greater weight on a unit of health gain for *non*-end of life patients than on that for end of life patients, *ceteris paribus.* This finding was unexpected, though not entirely unique (see, for example, Skedgel *et al.*, 2015). Some efforts were made to understand this type of preference, by way of probing debrief questions in empirical study 1, tick-box tasks to capture the reasons for respondents' choices in empirical study 2, and attitudinal questions to test for internal coherence of responses in empirical study 4. The open-ended questions (as used in empirical study 1) were generally more insightful than the closed-ended questions (as used in empirical studies 2 and 4), but none of the efforts proved to be particularly enlightening. It was found that general public respondents are often unable to articulate the reasons for their choices, and that their responses are often inconsistent. Hence, a limitation of the research is that the *reasons* why people would support an end of life 'penalty' (as many appear to) remains unexplained.

In the discussion of the literature review (see 2.4.2), it was contended – based on evidence reported by Shah *et al.* (2015b) – that respondents who choose to treat a non-end of life patient rather than an end of life patient (when the gains to both patients are the same) may be doing so based on an expectation that other respondents will choose to treat the end of life patient. Hence, choosing to treat the non-end of life patient is a means of increasing the likelihood that the overall outcome of the study will be neutral, which may well be the respondent's true preference. A similar argument can be applied to the observation of respondents making choices that are supposedly dominated (see 7.4.1). Although it is probable that respondents making such choices (particularly when an indifference option is available) are doing so because they have failed to understand or pay sufficient

attention to the task, it is possible that they simply reject the provision of health care based on expected treatment gains, and they have judged that choosing the dominated option is a means of increasing the likelihood that the overall study result will be that of equal priority to both patients. Strategic bias and tactical responding have been discussed in the contingent valuation literature (Milon, 1989; Tilling *et al.*, 2016) but rarely in relation to health care priority-setting and empirical ethics.

A more investigative approach to understanding and interpreting respondents' stated preferences and choice strategies, based on qualitative research methods, would have been beneficial. In a recent survey of authors of health care-related DCE studies, all of the researchers surveyed who had reported using qualitative methods stated that these methods had added value to their choice experiments (Vass *et al.*, 2017). However, qualitative data can be difficult and resource-intensive to analyse, particularly in large sample studies. One option could be to invite selected respondents (either a random selection or those who gave responses of particular interest) to attend an in-depth debrief interview or focus group discussion. It is acknowledged that there may be practical issues associated with this kind of approach. Panel providers may not typically permit face-to-face contact with their members, and even if they did, there is likely to be selection bias as those agreeing to take part in follow-up activities may not be representative of the panel membership. Further, test-retest reliability may be low at the individual level for priority-setting preference studies, which would make it difficult to develop meaningful debrief questions. Nevertheless, it would be worth exploring the feasibility of using qualitative methods to acquire additional information about preferences and considering whether the aforementioned challenges can be overcome.

There is interest in the field of health state valuation in giving respondents completing stated preference surveys the opportunity to deliberate and reflect on their views, in order to ensure that the resulting choice data are a meaningful, carefully considered reflection of their preferences (Robinson and Bryan, 2013; Karimi *et al.*, 2016; Shah, 2016b). This type of approach is informed by the view that people lack clearly formulated preferences for most evaluation tasks (Fischoff, 1991) and are constructing their preferences on the spot in response to the particular tasks and question frames they are being presented with (as opposed to the view that people have pre-existing, consistent and stable values that researchers can 'tap into') (Dolan, 1999). Such approaches may be helpful in the context of eliciting end of life-related preferences, particularly to understand whether inconsistencies and ambiguities in respondents' choices are caused by their

(mis)interpretation of the choice task or in fact reflect their considered preferences. It has been shown elsewhere that people's views about priority-setting can change when they are given the opportunity to deliberate and discuss the issues (Dolan *et al.*, 1999). It is conjectured that deliberative methods can also promote the articulation of ideas and allow a wider range of rationales to emerge.

7.5.1.2 Range of life expectancies presented

The focus on NICE's policy resulted in a specific definition of end of life – that is, a life expectancy of less than two years. It is acknowledged that this cut-off is arbitrary and may not be relevant in other settings. All four of the empirical studies presented a relatively narrow range of life expectancy levels (albeit fairly consistent with the rest of the literature; see Appendix 3), often comparing a patient who would meet NICE's criteria with another patient who would not, but whose life expectancy was still very short by most people's standards. The methods are therefore relevant to understanding whether NICE's life expectancy criterion is supported, but may be insufficient for determining the extent of support for a more general policy of prioritising on the basis of life expectancy. Further research should extend the range of life expectancies presented, in order to understand whether the preferences observed with regard to prioritising the patient with shorter life expectancy are generalisable. It is plausible, for example, that a respondent would be indifferent between treating a patient with one year left to live and treating another patient with three years left to live, but would prioritise a patient with 10 years left to live over another patient with 30 years left to live. Any such research would need to account for potential confounding effects of age-related preferences. Another avenue of research could be use preliminary questions to identify what respondents consider to be 'end of life' and to use their responses to direct them to a selection of choice tasks that are pertinent to their views.

7.5.1.3 Comparators

A related issue is that when claims are made that research findings support or do not support an end of life premium, it is reasonable to ask what end of life is being compared to (Pinto-Prades *et al.*, 2014). As noted in 7.3.1, the empirical studies in this thesis focused on comparing short life expectancies to longer life expectancies. This is in line with the majority of the literature – most of the studies included in the literature review were similarly limited in their choice of comparators (see 2.3.7). Future studies should also elicit preferences for prioritising end of life treatments over other comparators, such as preventive interventions (Gyrd-Hansen, 2017), treatments for chronic conditions, and treatments to alleviate temporary losses of quality of life. Such comparators are relevant because under

the assumption of a single, fixed health care budget, relaxing the funding requirements for end of life treatments means raising the requirements for *all* other types of treatments.

7.5.1.4 Use of indifference options and visual aids

Further research is recommended on the use of indifference options in stated preference studies. This thesis has provided evidence that respondents' choices were influenced by the availability of an explicit indifference option, and by the wording of that option. However, the range of indifference options examined was limited, which restricts the generalisability of the findings. Further research could compare the indifference options used in empirical study 4 with alternatives such as 'don't know' options, equal splits of resources (as used by Green (2009) and Linley and Hughes (2013)), or options designed to reflect a desire to avoid making difficult decisions (for example, 'I am not able to make a decision and would prefer that the choice be made by others', also used by Green (2009)). Such research could investigate whether respondents consider these alternative options – which may well have similar or identical implications for the allocation of health care resources – to mean the same or different things.

This thesis has also been provided evidence that the propensity to express indifference in a given choice task can depend on *when* in the survey that task appeared (see 6.3.10), though no attempt was made to examine whether this propensity is affected by the *nature* of the preceding tasks. This could be explored in further research. For example, if the survey were to begin with a choice involving a pair of dominant/dominated options, it is hypothesised – based on existing evidence of anchoring effects (Tversky and Kahneman, 1992) – that respondents would be less likely to express indifference in subsequent tasks than if the survey were to begin with a more 'difficult' choice. In a similar vein, the impact of combining forced choice and non-forced choice designs could be investigated. This might involve starting each task as a forced choice, then making a third option of indifference available once respondents had made their initial choice. This could be compared to a test arm that includes an indifference option throughout, or one that starts with an indifference option before reverting to a forced choice for respondents who initially express indifference. It is hypothesised that respondents will be less willing to express indifference after having committed to choosing one of the forced choice options than if an indifference option had been available throughout.

A study could also be undertaken to examine the impact of mode of administration on the propensity to express indifference – specifically, to test the hypothesis (as

suggested in 7.3.4) that respondents are more inclined to choose an indifference option in internet surveys that in in interviewer-led modes of administration. This research question cannot be answered with recourse to the existing literature on end of life-related preferences - neither of the studies that used multiple modes of administration (Richardson *et al.*, 2012; Rowen *et al.*, 2016b) included an indifference option.

Just as the use of indifference options could be subjected to further investigation, there is also scope for extending the examination of the use of visual aids in health care priority-setting studies. The results of empirical study 4 suggested that the use of graphical presentations of information affected respondents' choices in some scenarios but not in others. However, a limitation is that only one type of visual aid was used – a conceptual diagram that was near-identical to those used in empirical study 3 and the two Rowen *et al.* studies. Further research could compare different forms of visual aid, not only in terms of what choice data they generate but also in terms of the ways in which respondents perceive and interpret the information contained in the graphics. Van de Wetering *et al.* (2015) propose the use of think-aloud procedures for this purpose. It has also been suggested that moving pictures (for example, animated graphs that show how health changes over time) and physical props (such as the wooden boards traditionally used in time trade-off interviews) can help improve respondent engagement and understanding (Lo, 2017).

It is acknowledged that these kinds of methodological experiments have the potential to complicate matters and make it more challenging for researchers to draw unequivocal conclusions from stated preference studies. However, given the known importance of framing effects, it is helpful to be able to understand whether the preferences elicited in a given study are stable or if they are likely to be an artefact of the study design (Ubel, 1999). The findings of such experiments can also provide guidance to researchers who are considering whether and how to include indifference options, visual aids and other features in their own studies, and what effect their chosen approach is likely to have on the resulting data relative to other candidate approaches.

7.5.1.5 Preferences for quality of life-improving treatments

The finding (common to empirical studies 1, 2 and 4) that quality of life improvements are favoured to equal-sized (in QALY terms) life extensions in an end of life context was obscured somewhat by the finding in empirical study 4 that similar preferences were also observed in a non-end of life context. This suggests that many respondents may have been expressing a social value judgement that

quality of life-improving QALYs should be prioritised over life-extending QALYs, even if both types of QALYs are equally valuable (in individual utility terms) to patients themselves.[25] It could, however, call into question whether respondents assumed (as had been intended) that a quality of life improvement worth half a QALY would be considered equally desirable to the patients as a life extension worth half a QALY. It is plausible that respondents had ignored, rejected or failed to understand the implications of the instruction provided about how to interpret the concept of 50% health, instead assuming that the patients would in fact prefer the quality of life improvement for themselves.

Disentangling individual and social values is not straightforward. One possible avenue of research would be to identify, for each individual respondent, a specific health state – defined using a preference-based measure such as EQ-5D – deemed by that respondent to have a value of 0.5 from the perspective of their own health. This would involve the respondent expressing indifference between living in that state for n years and living in full health for n/2 years. The time trade-off technique, which involves trading length of life for improved quality of life, would be well suited for this purpose, though some effort would be required to find a health state with a value sufficiently close to 0.5. This might involve first conducting a time trade-off valuation of a given EQ-5D health state. Then, depending on whether that health state was valued higher or lower than 0.5, the task would need to be repeated using a different health state slightly worse than or slightly better than the previous health state (by adjusting the level of one of the dimensions). Once a health state deemed to represent 0.5 has been identified, the same respondent could be presented with social decision-maker perspective tasks similar to those used in scenarios S4 and S6 in empirical study 4, but with 50% health defined specifically in terms of the EQ-5D health state that they themselves had valued at 0.5. An alternative approach – which avoids the difficulty in aggregating data on preferences for different health states – would be to present the same health state to all respondents. The parameters of the subsequent social decision-maker perspective tasks would need to be adapted to account for the particular time trade-off value provided by each respondent.

7.5.1.6 Preference heterogeneity and choice strategies

The thesis reports a variety of analyses seeking to identify the characteristics of respondents who responded to choice tasks in certain ways. In empirical study 3, the background characteristics of respondents who consistently followed certain

[25] It should be noted that previous research has suggested that in non-end of life contexts, gains from life extensions are favoured over gains from quality of life enhancement, holding constant the size of the gains (Olsen and Donaldson, 1998; Pennington *et al.*, 2013; Gu *et al.*, 2015).

choice strategies of interest (such as always choosing to treat the patient with shorter life expectancy without treatment) were reported. Multinomial logit regressions were used to assess the extent to which these characteristics affected the likelihood of respondents belonging to a particular subgroup defined by their choice behaviour. In empirical study 4, a similar type of analysis was used to assess the impact of background characteristics on the likelihood of respondents choosing to give priority to the end of life patient in scenario S1. In both cases – and consistent with many of the studies identified in the literature review – the impact of the background characteristics was small, and respondents' choices were not found to be well predicted by the observable characteristics on which information was collected in the relevant studies. It is acknowledged that the analytical approach adopted was fairly limited and that more rigorous forms of cluster analysis which allow for unobserved heterogeneity in preferences – such as latent class analysis – might be better suited to identifying relevant subgroups.

7.5.1.7 Preference elicitation methods

A general drawback of the studies reported in this thesis – and of many other studies in the literature – is that they require general public survey respondents to consider several pieces of numerical information simultaneously, and often to make unassisted calculations using numerical data. The subsequent results reported generally assume that respondents – who may or may not have high levels of numeracy – have interpreted the data and made the necessary calculations correctly. For example, in scenario S3 of empirical study 4, respondents were expected to have understood that by the time of the patients' deaths in absence of treatment, patient A will have known about their prognosis for longer (and therefore will have had more time to prepare for death) than patient B. This is not self-evident and was not stated explicitly in the information presented. The piloting work did not indicate that this was problematic, but it is acknowledged that the pilot was not undertaken in similar conditions to the main study and no attempt was made to test the survey on a sample with below average levels of numeracy. Evidence of respondents in stated preference studies making responses that indicate misinterpretations of numerical information are reported both in this thesis (for example, see 4.3.1) and elsewhere (for example, see Veldwijk *et al.*, 2016). This issue may be overcome by an increasing role for research methods that generate preference data but are less reliant on respondents' numeracy, such as Q methodology. However, it should also be noted that such methods may be insufficient to generate QALY weights unless combined with other approaches. For example, having identified and described three perspectives on the value of end of life treatments using Q methodology in McHugh *et al.* (2015), the authors have

proceeded to investigate whether they can measure the distribution of those views across a large, representation sample using an alternative survey approach (Baker, 2016).

7.5.1.8 Reliability of the findings

The empirical studies shared similar aims, methods and designs, but they differed sufficiently from each other so as to obscure direct comparisons between the results of (a given question in) one study with those of (a corresponding question in) another. A limitation of the research, therefore, is that little attempt was made to include an identical question in multiple studies in order to assess inter-study reliability, or to administer the same survey to the same respondents twice to assess test-retest reliability. To the author's knowledge, such reliability tests are scarcely reported in the empirical ethics literature.

7.5.2 Ideas for future research beyond the scope of the thesis

7.5.2.1 Extending scope beyond the preferences of the UK general public

Given the focus on NICE, the scope of the research was limited to the preferences of the UK general public.[26] However, the question of whether end of life treatments should be subject to special weighting is not only of interest in the UK – the literature review showed that the issue has also been investigated in countries such as Norway (Abel Olsen, 2013) and Spain (Pinto-Prades *et al.*, 2014). The author is aware of a just-published paper reporting similar research in the Netherlands (Wouters *et al.*, 2017) and working papers reporting studies undertaken in Denmark (Gyrd-Hansen *et al.*, 2017) and Sweden (Olofsson *et al.*, 2016). It would be informative to apply the methods used in this thesis to elicit the preferences of non-UK populations, in order to examine whether the differences in findings observed between studies can be explained by differences in the setting (bearing in mind that Table 2-6 suggests that studies conducted in the UK report evidence consistent with an end of life premium less frequently than those conducted elsewhere).

Likewise, it would be informative to elicit the preferences of other respondent groups, such as individuals involved in NICE's technology appraisal processes and policy makers responsible for making prioritisation decisions on behalf of the public.

[26] It is worth noting that although the work reported in this thesis is independent research and the author maintained full control over all aspect of the work (including design, conduct and interpretation) at all times, financial support for all four empirical studies was provided by NICE via its Decision Support Unit. The author sought the feedback from selected NICE employees and advisors (see Acknowledgements), particularly during the early stages of the research programme, to ensure that the studies were designed so as to generate results that would be useful to NICE. Hence, although the focus on NICE can be described a limitation, to some extent it is by design.

This would provide insight into whether policy makers – who may be described as being part of an implicit principal-agent relationship (Coast, 2001) – make choices that reflect those of the public (Skedgel *et al.*, 2015). Such research would complement previous studies that used stated preference (Tappenden *et al.*, 2007) and revealed preference methods (Dakin *et al.*, 2015) to examine the influence of factors other than cost-effectiveness on recommendations made by NICE's appraisal committees.

7.5.2.2 Condition-specific preferences

All of the studies reported in this thesis used unlabelled designs in that the hypothetical patients were not described as having a particular named condition. This is in keeping with the rest of the empirical literature – 16 of the 20 studies included in the literature review did not specify the names of the conditions (Table 2-5). The use of generic labels (patient A and patient B) was guided by concerns that the use of condition labels would induce emotional and biased responses. This approach is consistent with NICE's end of life policy, which in principle does not distinguish between different illnesses or treatments. The generic presentation of health care priority-setting scenarios is supported by the findings of Roberts *et al.* (1999) who report that respondent engagement levels are not sensitive to the provision of supporting clinical information.

However, NICE's policy is sometimes interpreted as a 'cancer premium' since in practice only cancer drugs have met the criteria for special consideration (Collins and Latimer, 2013; McCabe *et al.*, 2016b). The existence of the CDF also suggests that there is deemed to be something 'special' about cancer. Yet end of life and cancer are not synonymous – a policy of giving extra weight to cancer (and only cancer) treatments would exclude other terminal or end of life conditions, such as motor neurone disease and advanced heart disease (Shah, 2017). Moreover, many cancer treatments are indicated for patients with early stage disease who might expect to live for much longer than the 24-month cut-off specified in NICE's end of life guidance (and may not die as a result of cancer).

The research in this thesis could be extended by exploring the interaction between preferences regarding end of life and preferences regarding cancer. It could be that even in unlabelled studies, people connect end of life scenarios with cancer, and the preferences they express may therefore reflect 'dread' effects associated with that groups of diseases.[27] The resulting policy question would then be whether it is

[27] Viscussi *et al*. (2014) describe cancer as a dread disease on the grounds that it generates fear that is disproportionate to the actual health impact and risks associated with the disease.

legitimate to base reimbursement decisions on the fears of the general public, which may be the product of biases and misconceptions (Shah, 2017).

7.6 Policy implications and concluding remarks

This thesis has sought to answer 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 – a research question that had been motivated by NICE's policy for appraising end of life treatments. The programme of empirical research undertaken has provided little indication that this is the case. While empirical studies 1 and 2 reported tentative evidence of support for prioritising the treatment of end of life patients, empirical studies 3 and 4 – which used larger samples and more robust methods – reported results that are largely inconsistent with an end of life premium. The findings reported in the wider literature have been mixed, though it should be noted that the studies conducted in the UK have not, on the whole, reported evidence consistent with an end of life premium (Table 2-6). This has relevance for assessing the legitimacy of NICE's end of life policy, which was assumed to have been motivated at least in part by the views held by the population that the NHS serves. The results of empirical studies 1, 2 and 4 also suggest that the focus on life-extending rather than on quality of life-improving treatments in NICE's end of life policy does not appear to be supported by the public either.

Based on these findings, it might be considered appropriate for NICE to abandon its end of life policy and any other mechanisms that relax the cost-effectiveness requirements for end of life treatments, on the grounds that the population health losses that arise due to the policy are not justified by the evidence on societal preferences. This would result in fewer approvals of end of life treatments, and therefore in reduced access to treatments for patients with terminal illness. Hence, some patients would lose out as a result, and the reigniting of issues that had instigated the initial introduction of NICE's end of life policy would be inevitable. In principle, however, other, less identifiable groups of patients would benefit as the freed funding could be spent on health care that is more cost-effective and/or that the public values more.

It may be that there are compelling arguments for retaining some form of end of life weighting irrespective of public preferences. For example, if the standard QALY approach used by NICE systematically underestimates the (health or non-health) benefits of end of life treatments (whether or not this is actually the case would itself need investigating), it may be appropriate to correct for this. An end of life premium may also help to encourage innovation, or to meet broader health system

and political objectives. Furthermore, once a prominent policy has been introduced, it may be inherently and procedurally difficult to withdraw that policy.

At the time of writing, the policy situation is that there are no imminent plans to proceed with the introduction of burden of illness weights as had been planned as part of the value-based assessment proposals. However, the approach continues to be considered by NICE and may be revived in the future. It is clear that there is overlap between an end of life premium and the concept of weighting QALYs based on severity and burden of illness (whether operationalised in terms of absolute QALY shortfall, proportional QALY shortfall, or some other measure). Weighting QALYs in accordance with a continuous variable that captures quality of life as well as life expectancy may be more consistent with public preferences than the current practice of applying binary cut-offs based only on life expectancy information, and would address some of the criticisms of the arbitrariness of NICE's end of life policy.

The thesis has also provided evidence that where public preferences consistent with an end of life premium *do* exist, these preferences may depend to some degree on how long the end of life patients have known about their prognosis. A pertinent question to ask is whether this is a relevant policy consideration – is it feasible that NICE would ever incorporate this factor into a revised version of its end of life policy? It is acknowledged that it is unrealistic to expect NICE to make one recommendation for end of life patients who have just learned about their prognosis and another for end of life patients who have known about their prognosis for some time (though it may be feasible to make separate recommendations for different indications and disease stages). Nevertheless, it is informative to understand the rationale behind people's preferences, both to help make sense of the results of stated preference studies and to determine whether a policy that purports to reflect public preferences would actually lead to outcomes that would enjoy widespread support.

In the extreme, suppose a state of the world in which all end of life treatments being considered by NICE target patients who have known about their prognosis for some time (for example, patients at the late stage of a slow-progressing terminal illness that invariably presents and get diagnosed early). In an alternative state of the world, suppose that all end of life treatments target patients who have only recently learned of their prognosis (for example, patients with a condition that invariably presents and gets diagnosed late). In the former case, if there is robust evidence that any observed public support for an end of life premium is driven by concerns about preparedness and not by concerns about short life expectancy *per se*, then this would imply that the end of life premium currently applied by NICE is

not consistent with public preferences. In the latter case, the end of life premium *would* receive public support because the treatments that would stand to receive special weighting would benefit patients with limited amounts of time to prepare for death.

Similar questions can be asked about the policy relevance of the finding in empirical study 4 that respondents' preferences regarding an end of life premium appear to depend on the ages of the affected patients. This is consistent with findings elsewhere in the literature that people tend to become less concerned about patients' remaining life years when those patients are relatively old (see 2.3.8.4). Although NICE cannot restrict access to health care on the basis of age unless age is a good indicator of clinical effectiveness, it is informative to know whether any observed support for an end of life premium is contingent on such characteristics. To give another hypothetical example: if many or all of the end of life treatments being considered by NICE happen to target older patients, and public support for an end of life premium exists but only for younger patients, it follows that the end of life premium currently applied by NICE would *not* be supported.

It is acknowledged that in practice the selective application of an end of life premium in certain circumstances or for certain patient groups may have unintended and undesirable implications. It may be that older end of life patients tend to share some other characteristic that younger end of life patients tend not to possess, so a policy that gives lower priority to treating older end of life patients would inadvertently result in hurting disproportionately those with that particular characteristic. This may be inequitable in itself, even before taking into account any possible interaction between people's end of life-related preferences and their preferences regarding that characteristic. For any future amendment of its end of life policy, NICE would need to consider carefully the identity of the potential winners and losers, and to assess the likely outcomes in relation to its principles on social value judgements about equity.

The empirical literature on the relative weighting of QALYs, of which the literature on end of life-related preferences examined in thesis is part, is rapidly growing. It is clear that NICE and other UK decision-making agencies are interested in public preferences to some extent. This is demonstrated by the existence of the NICE Citizens' Council; the claims made in official documents and unofficial papers authored by employees of and advisors to these agencies; and the fact that the agencies occasionally initiate and fund research on public preferences (including several of the studies discussed in this thesis). The extent to which this kind of

evidence actually influences decisions (and indeed, whether it *ought* to influence decisions) is less clear.

The author's view is that the empirical ethics process could be improved if stated preference researchers were to be given clearer signals from decision-makers about which topics they would like to see prioritised in applied research that seeks to inform policy. Decision-makers should also communicate clearly any concerns or doubts they have about existing research so that efforts can be made to address those concerns – for example, through improvements in methodology. Clearer communication of this sort would help make future empirical studies more useful and policy relevant. Subject to the decision-makers' requests being compatible with research ethics and intellectual curiosity, most researchers would likely relish the opportunity to collaborate and engage in the co-production of knowledge that will ultimately be of benefit of society.

APPENDICES

Appendix 1 List of acronyms and glossary

List of acronyms

CDF	Cancer Drugs Fund
DCE	Discrete choice experiment
EQ-5D	EuroQol five-dimension instrument
ICER	Incremental cost-effectiveness ratio
LE	Life expectancy
NHS	National Health Service
NICE	National Institute for Health and Care Excellence
PTO	Person trade-off
QALY	Quality-adjusted life year
QOL	Quality of life
SSCI	Social Sciences Citation Index
UK	United Kingdom
WTP	Willingness-to-pay

Glossary

A selection of concepts used regularly in the thesis are explained below. It is acknowledged that some of these definitions are restrictive or simplistic, and that alternative definitions for these concepts exist. The intention is not to provide a comprehensive review of concepts but to briefly summarise the specific ways in which a selection of key concepts have been defined and interpreted in this thesis.

Appraisal / health technology appraisal

Within the context of NICE's technology appraisal programme, this is the process of making recommendations about the use of new and existing technologies in the NHS. These recommendations are guided by evidence of the technologies' effectiveness and cost-effectiveness; and by scientific and social value judgements made by NICE's advisory committees.

Budget allocation

A stated preference technique which asks survey respondents to indicate how a fixed budget should be allocated between two (or more) competing programmes.

Choice-based method

A method that requires a trade-off to be made (Whitty *et al.*, 2014). Person trade-off and willingness-to-pay are examples of choice-based methods. These can be compared to non-choice-based methods such as opinion polls and rating exercises.

Discrete choice experiment

A stated preference technique which asks survey respondents to make choices between two or more discrete alternatives where at least one attribute of the alternatives is systematically varied in such a way that information related to preference parameters of an indirect utility function can be inferred (Carson and Louviere, 2011).

Efficiency

Either the maximisation of output or benefit for a given cost, or the minimisation of cost to achieve a given output or benefit. An example of an efficiency objective is to seek to maximise population health using available resources.

Equity

Related to ethical judgements about the fairness of distributions and the fair treatment of individuals. Horizontal equity refers to the equal treatment of equals – a distribution is said to be horizontally equitable when people are treated the same in some relevant respect. Vertical equity refers to the unequal treatment of unequals – a distribution is said to be vertically equitable when people who are different in some relevant way are treated appropriately differently (for example, according more of some relevant entity to those in greater need of it) (Culyer, 2010).

Framing effect

An example of cognitive bias whereby people's reaction to a given choice is influenced by the way is which that choice is presented (Plous, 1993).

Full health

A state involving no morbidity problems, thereby assigned a quality of life weight of one. This is consistent with the label of the upper anchor used in EQ-5D valuation studies (Shah *et al.*, 2016).

Health

A combination of quality of life (morbidity) and length of life (longevity), as measured using quality-adjusted life years. Also used to describe quality of life (distinct from length of life) in the surveys used in the empirical studies in this thesis – see below.

Health care

Goods and services provided to promote health or to prevent, alleviate or eliminate ill health (Culyer, 2010).

Health technology

Any method for promoting health or preventing/postponing ill health (Culyer, 2010), including but not limited to drugs. NICE (2017b) uses the term to mean a drug or other treatment that is being assessed. In this thesis, the NICE definition is adopted, and the terms 'technology', 'intervention' and 'treatment' are used interchangeably.

Incremental cost-effectiveness ratio

The ratio of the difference in costs between two alternatives to the difference in effectiveness between the same two alternatives (Brazier *et al.*, 2017).

Indifference

The situation where the utility gained by an individual from either of two entities is the same (Culyer, 2010).

Person trade-off

A stated preference technique which asks respondents how many outcomes of one kind they consider to be equivalent in social value to X outcomes of another kind (Nord, 1995).

Q method

A research method that combines qualitative and quantitative techniques to study people's 'subjectivity' (that is, their subjective opinions, values or beliefs). These methods enable the identification and description of shared views around a given topic (McHugh *et al.*, 2015).

Quality-adjusted life year

A generic measure of health that combines quality of life and length of life in a single index (Weinstein *et al.*, 2009). One quality-adjusted life year is equivalent to one year of life in full quality of life.

Quality of life

The morbidity aspect of health. Someone in full quality of life can be said to have no morbidity problems. This can be compared to broader uses of the term, referring to a construct reflecting subjective or objective judgement concerning all aspects of an individual's existence, including not only health but also economic, political, cultural, environmental, aesthetic and spiritual aspects (Brazier *et al.*, 2017). Numeric measurements of quality of life can be used to weight numeric measurements of length of life in the calculation of quality-adjusted life years. For this purpose, quality of life is measured on scale that is anchored at one (representing no morbidity problems) and zero (representing dead). The terms 'health-related quality of life' and 'health status' are often used to describe the same concept (Karimi and Brazier, 2016) but these terms are not used in this thesis. In the surveys used in the empirical studies in this thesis, the terms 'health' and 'general health' were used to distinguish quality of life from length of life. This was because of concerns that the term 'quality of life' would have been interpreted by respondents in terms of the broader construct described above.

Social grade

A system of demographic classification based on the occupation of the head of the household, originally developed by the National Readership Survey.

Social preference

A preference concerning the allocation of resources for others in society. In this thesis, the terms 'social preference', 'societal preference', 'social value' and 'societal value' are used interchangeably. NICE's Citizens' Council favours the term 'societal values' (NICE, 2014b).

Stated preference

Willingness to engage in trade-offs to acquire a good, service or non-marketed entity as derived from questionnaires or experiments. The preference is stated verbally or numerically rather than revealed by actual behaviour in experiments or in real life (Culyer, 2010).

Trade-off

The notion of voluntarily sacrificing some of one good in exchange for a sufficient increase in the amount of some other good (Culyer, 2010).

Utility

The preference for, or desirability of, a particular outcome (Brazier *et al.*, 2017).

Welfarism / extra-welfarism

The main school of thought within modern welfare economics that holds that judgements of social welfare must be a function of individual utility, as judged by the individuals themselves (Brazier *et al.*, 2017). By comparison, non-welfarism holds that judgements of social welfare can be based on information other than individual utility. One form of non-welfarism – extra-welfarism – commonly postulates health itself as the maximand of the health care sector, rather than the individual utility to which it may give rise (Culyer, 2010).

Willingness-to-pay

The maximum sum an individual or government is willing to pay to acquire some good or service, or to avoid a prospective loss. Willingness-to-pay can be elicited from stated or revealed preference experiments (Culyer, 2010).

Without treatment

Refers to the situation when the treatment on offer is not received and the benefits it generates are therefore not realised. Strictly speaking, the patient(s) failing to receive the treatment would receive best supportive care rather than no treatment at all.

Appendix 2 Results of preliminary SSCI searches

All of the search strategies below were tested in May 2014.

Table 0-1. Initial search terms

Search	Results
TS=("end of life")	6,551
TS=preferences	82,537
TS=("end of life") AND TS=preferences	1,076

Table 0-2. Addition of terms related to end of life

Search	Results
TS=("end of life") AND TS=preferences	1,076
TS=severity AND TS=preferences	837
TS=terminal AND TS=preferences	655
TS=("end of life" OR severity OR terminal) AND TS=preferences	2,387

Table 0-3. Addition of terms identified in key paper abstracts

Search	Results
TS=("end of life" OR severity OR terminal) AND TS=preferences	2,387
TS=("end of life" OR severity OR terminal) AND TS=preferences AND TS=health	1,022
TS=("end of life" OR severity OR terminal) AND TS=(preferences) AND TS=health AND TS=(respondents OR subjects OR participants OR sampl*)	539

Table 0-4. Testing addition of further terms identified in key paper abstracts

Search	Results
TS=("end of life" OR severity OR terminal) AND TS=(preferences) AND TS=health AND TS=(respondents OR subjects OR participants OR sampl*)	539
TS=("end of life" OR severity OR terminal) AND TS=(preferences) AND TS=health AND TS=(respondents OR subjects OR participants OR sampl*) AND TS=(patients OR treatments)	439
TS=("end of life" OR severity OR terminal) AND TS=(preferences) AND TS=health AND TS=(respondents OR subjects OR participants OR sampl*) AND TS=(public OR popul*)	185
TS=("end of life" OR severity OR terminal) AND TS=(preferences) AND TS=health AND TS=(respondents OR subjects OR participants OR sampl*) AND TS=(evidence)	56

Table 0-5. Addition of further term related to end of life

Search	Results
TS=("end of life" OR severity OR terminal) AND TS=(preferences) AND TS=health AND TS=(respondents OR subjects OR participants OR sampl*)	539
TS=("end of life" OR severity OR terminal OR "life expectancy") AND TS=(preferences) AND TS=health AND TS=(respondents OR subjects OR participants OR sampl*)	598

Appendix 3 Tabular summary of included studies

Note: The fields in this table are the same as those in Table 2-1.

Record	**Abel Olsen (2013)**
Year of publication	2013
Year of study conduct	2010
Country or countries of origin of data	Norway
Sample size	503
Type of sample	Public
Sample recruitment process	Recruited by agency
Criteria for excluding respondents and/or observations reported?	No
Mode of administration	Internet survey
Summary of primary study objective(s)	To test for support for end of life prioritisation and the fair innings approach
Was end of life (or a related term) mentioned explicitly in the study objectives?	Yes
Pilot reported?	No
Preference elicitation technique	Other choice exercise
Perspective	Social decision-maker
End of life definition	Remaining lifetime without treatment
Life expectancy without treatment attribute levels	1yr, 3yrs, 10yrs
Life expectancy gain from treatment attribute levels	1mth, 3mths, 1yr, 3yrs
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No
What were respondents choosing between (or choosing to do)?	Which of two patients to treat

Record	**Abel Olsen (2013)**
Was it possible to express indifference?	Yes – following an initial forced choice without an indifference option, respondents were asked to specify how large a gain their less preferred patient would need in order for the two patients to have equal priority (hence, although respondents were never given an explicit indifference option to choose, they were able to express indifference by specifying a size of gain for their less preferred patient that was no different from that indicated in the initial forced choice question)
Were visual aids used?	No
Strength of preference examined at the individual respondent level?	Yes – using benefit trade-off type approach
Number of tasks completed by each respondent	4
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Evidence not consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	Evidence of support for the fair innings approach
Other factors examined	Fair innings approach, health gain
Impact of background characteristics reported?	No
Were qualitative data or explanatory factors sought?	Yes – respondents were asked which factor was most important to them when answering the questions
Was any reference made to age-related preferences?	Yes – evidence of a desire to reduce inequalities in age at death
Was any reference made to time-related preferences?	No

Record	**Baker *et al.* (2010a)**
Year of publication	2010
Year of study conduct	2007
Country or countries of origin of data	UK
Sample size	587
Type of sample	Public
Sample recruitment process	Recruited by agency
Criteria for excluding respondents and/or observations reported?	No
Mode of administration	Computer-assisted personal interview
Summary of primary study objective(s)	To test for support for multiple prioritisation criteria
Was end of life (or a related term) mentioned explicitly in the study objectives?	No
Pilot reported?	Yes
Preference elicitation technique	Discrete choice experiment
Perspective	Social decision-maker
End of life definition	Age at onset and age at death if untreated were included as variables; when age at onset = age at death if untreated, the profile describes an imminent death scenario where any treatment is life-saving/extending
Life expectancy without treatment attribute levels	0yrs, 9yrs, 10yrs, various levels > 10yrs (not presented explicitly, but can be calculated indirectly by subtracting age at onset from age at death if untreated)
Life expectancy gain from treatment attribute levels	0yrs, 1yr, 5yrs, 10yrs, 20yrs, 40yrs, 60yrs, 79yrs
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No – when age at onset = age at death if untreated, all treatments are necessarily life-extending
What were respondents choosing between (or choosing to do)?	Which of two patient groups to treat

Record	**Baker *et al.* (2010a)**
Was it possible to express indifference?	No
Were visual aids used?	Yes
Strength of preference examined at the individual respondent level?	No
Number of tasks completed by each respondent	8 (+6 tasks using a different method that did not examine end of life, as well as attitudinal questions)
Time taken to complete survey reported?	Yes – 41 min (average)
Summary of finding: end of life vs. non-end of life	Mixed or inconclusive evidence
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	In ranking exercise (n=19) conducted in preliminary study, life expectancy without treatment was ranked third out of 10 priority-setting attributes (below quality of life without treatment but above all patient characteristics, e.g. age, lifestyle); age and severity did not have a strong impact on choices over and above QALY gains
Other factors examined	Age at onset, age at death, life expectancy gain, quality of life without treatment, quality of life gain
Impact of background characteristics reported?	No
Were qualitative data or explanatory factors sought?	Yes – in preliminary work (but end of life was not a specific topic of discussion)
Was any reference made to age-related preferences?	Yes – evidence of preference for life-saving treatments for 10 year old patients but not for other patients of other ages
Was any reference made to time-related preferences?	No

Record	Dolan and Cookson (2000)
Year of publication	2000
Year of study conduct	Not reported
Country or countries of origin of data	UK
Sample size	60
Type of sample	Public
Sample recruitment process	Random postal invitations
Criteria for excluding respondents and/or observations reported?	No exclusions
Mode of administration	Focus group
Summary of primary study objective(s)	Qualitative examination of support for multiple prioritisation criteria
Was end of life (or a related term) mentioned explicitly in the study objectives?	No
Pilot reported?	No
Preference elicitation technique	Other choice exercise
Perspective	Social decision-maker (operationalised using a veil of ignorance condition for half of the respondents)
End of life definition	Life expectancy without treatment
Life expectancy without treatment attribute levels	0yrs, 5yrs, 10yrs, 30yrs (but the sole end of life vs. non-end of life question involved a choice between patients with life expectancies of 10yrs and 30yrs, respectively)
Life expectancy gain from treatment attribute levels	5yrs, 10yrs, 15yrs, 20yrs (but in the sole end of life vs. non-end of life question, life expectancy gain was 10yrs for both candidate recipient groups)
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No – quality of life was examined but in separate questions
What were respondents choosing between (or choosing to do)?	Which of two patient groups to treat (with the gain attribute then increased/reduced incrementally)

Record	Dolan and Cookson (2000)
Was it possible to express indifference?	Yes – a 'same priority' option was available
Were visual aids used?	No
Strength of preference examined at the individual respondent level?	Yes – attribute levels were varied incrementally
Number of tasks completed by each respondent	6 (+initial discussion and questionnaire on health care priority-setting in general)
Time taken to complete survey reported?	Yes – meeting lasted for two hours
Summary of finding: end of life vs. non-end of life	Mixed or inconclusive evidence
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	Veil of ignorance perspective (vs. social decision-maker) had no discernible impact; authors conclude from data that "equality of access should prevail over the maximisation of benefits" (p.19)
Other factors examined	Quality of life without treatment, quality of life with treatment; other factors were mentioned by respondents but these were either irrelevant or factors that they were not supposed to have considered (e.g. costs)
Impact of background characteristics reported?	Yes – none found to influence propensity to favour treating end of life patients
Were qualitative data or explanatory factors sought?	Yes – study was predominantly a qualitative exercise
Was any reference made to age-related preferences?	Yes – but age was intended to be an irrelevant factor
Was any reference made to time-related preferences?	No

Record	Dolan and Shaw (2004)
Year of publication	2004
Year of study conduct	Not reported
Country or countries of origin of data	UK
Sample size	23
Type of sample	Public
Sample recruitment process	Recruited by agency
Criteria for excluding respondents and/or observations reported?	No
Mode of administration	Focus group
Summary of primary study objective(s)	To test for support for multiple prioritisation criteria
Was end of life (or a related term) mentioned explicitly in the study objectives?	No
Pilot reported?	No
Preference elicitation technique	Other choice exercise
Perspective	Social decision-maker
End of life definition	Life expectancy without transplant
Life expectancy without treatment attribute levels	1yr, 4yrs, 7yrs, 10yrs, 13yrs, 16yrs
Life expectancy gain from treatment attribute levels	5yrs, 10yrs, 15yrs, 20yrs, 25yrs, 30yrs
Was disease labelled or named?	Yes – kidney failure
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No
What were respondents choosing between (or choosing to do)?	Which of six patients should receive a kidney transplant
Was it possible to express indifference?	No
Were visual aids used?	No

Record	Dolan and Shaw (2004)
Strength of preference examined at the individual respondent level?	No
Number of tasks completed by each respondent	3
Time taken to complete survey reported?	Yes – two meetings, each of which lasted for two hours
Summary of finding: end of life vs. non-end of life	Evidence not consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	Benefit from transplantation was the most important criterion overall; some participants chose to prioritise those with dependants
Other factors examined	Other factors mentioned by participants: age, family responsibilities, waiting time, cause, whether a re-transplantation or not
Impact of background characteristics reported?	No
Were qualitative data or explanatory factors sought?	Yes – study was predominantly a qualitative exercise
Was any reference made to age-related preferences?	Yes – the participants who had chosen to treat the patient with shortest life expectancy without transplant did not continue to do so when it was revealed that this patient was the oldest of the six candidate recipients
Was any reference made to time-related preferences?	No

Record	Dolan and Tsuchiya (2005)
Year of publication	2005
Year of study conduct	2002
Country or countries of origin of data	UK
Sample size	100
Type of sample	Public
Sample recruitment process	Postal invitation
Criteria for excluding respondents and/or observations reported?	Yes – excluded respondents who did not complete all of the tasks
Mode of administration	Self-completion paper survey (administered in group setting)
Summary of primary study objective(s)	To compare support for prioritisation according to age vs. prioritisation according to severity/life expectancy
Was end of life (or a related term) mentioned explicitly in the study objectives?	Yes
Pilot reported?	No
Preference elicitation technique	Ranking exercise and other choice exercise
Perspective	Social decision-maker
End of life definition	Future years without treatment
Life expectancy without treatment attribute levels	1yr, 6yrs
Life expectancy gain from treatment attribute levels	3yrs
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No – a question examining quality of life improvement was included, but the size of the life extension was fixed
What were respondents choosing between (or choosing to do)?	Which of six patient groups to treat; then to rank the six patient groups
Was it possible to express indifference?	No
Were visual aids used?	No

Record	Dolan and Tsuchiya (2005)
Strength of preference examined at the individual respondent level?	No
Number of tasks completed by each respondent	4
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Evidence not consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	Future health (quality of life without treatment) did not have a statistically significant effect on choices made, whereas past years (age) had a strong effect
Other factors examined	Past age, past health, quality of life without treatment
Impact of background characteristics reported?	Yes – age, education and employment status were all found to have statistically significant interactions with life expectancy without treatment
Were qualitative data or explanatory factors sought?	No – discussions were not recorded as it was not intended to be a qualitative study
Was any reference made to age-related preferences?	Yes – younger patient groups were always chosen over older ones
Was any reference made to time-related preferences?	No

Record	**Lim *et al.* (2012)**
Year of publication	2012
Year of study conduct	2010
Country or countries of origin of data	Korea
Sample size	800
Type of sample	Public
Sample recruitment process	Recruited by agency
Criteria for excluding respondents and/or observations reported?	Yes – excluded multiple responses from the same IP address
Mode of administration	Internet survey
Summary of primary study objective(s)	To test for support for multiple prioritisation criteria
Was end of life (or a related term) mentioned explicitly in the study objectives?	No
Pilot reported?	No (but focus groups were conducted, in part to inform the selection of attributes in the internet survey)
Preference elicitation technique	Discrete choice experiment
Perspective	Social decision-maker
End of life definition	Life expectancy without treatment
Life expectancy without treatment attribute levels	1yr, 5yrs, 10yrs, 20yrs, 25yrs, 35yrs
Life expectancy gain from treatment attribute levels	0yrs, 1yr, 5yrs, 10yrs, 20yrs, 30yrs
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No
What were respondents choosing between (or choosing to do)?	Which of two patients to treat
Was it possible to express indifference?	Not reported

Record	**Lim *et al.* (2012)**
Were visual aids used?	Yes
Strength of preference examined at the individual respondent level?	No
Number of tasks completed by each respondent	17
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Evidence consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	All attributes had statistically significant coefficients with signs that were consistent with the authors' expectations (QALY gain – positive; quality of life before treatment – negative; patient's household income – negative)
Other factors examined	QALY gain; quality of life before treatment; household income group
Impact of background characteristics reported?	No
Were qualitative data or explanatory factors sought?	Yes – using focus groups (but end of life was not a specific topic for discussion)
Was any reference made to age-related preferences?	No
Was any reference made to time-related preferences?	No

Record	**Linley and Hughes (2013)**
Year of publication	2013
Year of study conduct	2011
Country or countries of origin of data	UK
Sample size	4,118
Type of sample	Public
Sample recruitment process	Recruited by agency
Criteria for excluding respondents and/or observations reported?	No
Mode of administration	Internet survey
Summary of primary study objective(s)	To test for support for multiple prioritisation criteria
Was end of life (or a related term) mentioned explicitly in the study objectives?	No – end of life was one of many prioritisation criteria examined
Pilot reported?	Yes
Preference elicitation technique	Budget allocation
Perspective	Social decision-maker
End of life definition	Fatal disease that leads to death in 18 months without treatment
Life expectancy without treatment attribute levels	18mths, 60mths
Life expectancy gain from treatment attribute levels	3mths, 6mths
Was disease labelled or named?	No (but preferences regarding 'fatal cancer' were examined in a separate question)
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No
What were respondents choosing between (or choosing to do)?	To allocate a fixed budget between two groups of patients
Was it possible to express indifference?	Yes – a 50:50 split option was available

Record	Linley and Hughes (2013)
Were visual aids used?	No
Strength of preference examined at the individual respondent level?	Yes – respondents could choose from 11 different distributions of funding, and further 'health gain trade-off' and 'cost trade-off' approaches were also used
Number of tasks completed by each respondent	18 (of which two examined end of life explicitly)
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Evidence not consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	Evidence of support for prioritising treatment of severe illness, but not for prioritising treatment of cancer specifically
Other factors examined	Health gain; many others examined separately from end of life
Impact of background characteristics reported?	Yes – none found to influence propensity to favour treating end of life patients
Were qualitative data or explanatory factors sought?	No / not reported
Was any reference made to age-related preferences?	Yes – respondents did not support giving priority to the treatment of children overall (questions about children were separate from those about end of life)
Was any reference made to time-related preferences?	No

Record	**McHugh *et al.* (2015)**
Year of publication	2015
Year of study conduct	Not reported
Country or countries of origin of data	UK
Sample size	61
Type of sample	Individuals with different types of experiences or expertise in end of life in a professional or personal capacity (e.g. researchers, clinicians, people with experience of terminal illness)
Sample recruitment process	Purposive (to identify data-rich respondents)
Criteria for excluding respondents and/or observations reported?	No
Mode of administration	Non-computer-assisted personal interview
Summary of primary study objective(s)	Qualitative examination of societal perspectives in relation to end of life prioritisation
Was end of life (or a related term) mentioned explicitly in the study objectives?	Yes
Pilot reported?	Yes
Preference elicitation technique	Q methodology (technique that combines card sort and ranking exercise)
Perspective	Social decision-maker (though a minority of statements were framed using an own health perspective)
End of life definition	Described in multiple ways (e.g. 'terminally ill', 'die soon')
Life expectancy without treatment attribute levels	N/A
Life expectancy gain from treatment attribute levels	N/A
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	Yes
What were respondents choosing between (or choosing to do)?	Rank statements according to how much they agreed or disagreed with them

Record	**McHugh *et al.* (2015)**
Was it possible to express indifference?	Yes – in the grid ranging from -5 (most disagree) to +5 (most agree), respondents were able to place statements in the position marked 0
Were visual aids used?	No
Strength of preference examined at the individual respondent level?	Yes – strength of preference indicated by position in which statements were placed on the grid
Number of tasks completed by each respondent	1 (comprising sorting and placing of 49 statements on grid)
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Mixed or inconclusive evidence
Summary of finding: quality of life improvement vs. life extension	Mixed or inconclusive evidence
Other results of potential interest	Three shared accounts identified: (1) A population perspective – value for money, no special cases; (2) Life is precious – valuing life-extensions and patient choice; (3) Valuing wider benefits and opportunity cost – the quality of life and death
Other factors examined	Alternative perspectives and approaches to resource allocation – e.g. health-maximisation, provision of treatments to patients with non-terminal conditions
Impact of background characteristics reported?	Yes (though authors warn about making generalisations based on qualitative samples) – e.g. no academics helped to define the shared account most closely related to an end of life premium
Were qualitative data or explanatory factors sought?	Yes – study was in part a qualitative exercise
Was any reference made to age-related preferences?	Yes – e.g. one statement was worded: "I think life-extending treatments for people who are terminally ill are of less value as people get older"
Was any reference made to time-related preferences?	No reference to time discounting *per se*, but several statements referred to the value of time – e.g. "It is important to give a dying person and their family time to prepare for their death, put their affairs in order, make peace and say goodbyes"

Record	**Pennington *et al.* (2015)**
Year of publication	2015
Year of study conduct	2009-2010
Country or countries of origin of data	Denmark, France, Hungary, Netherlands, Norway, Poland, Spain, Sweden, UK
Sample size	17,657
Type of sample	Public
Sample recruitment process	Internet panel
Criteria for excluding respondents and/or observations reported?	Yes – excluded protest responders; respondents who expected to live for less than 6yrs were directed to a different questionnaire (not reported); impact of other exclusions reported in sensitivity analysis
Mode of administration	Internet survey
Summary of primary study objective(s)	To compare willingness-to-pay for different types of QALY gain
Was end of life (or a related term) mentioned explicitly in the study objectives?	No
Pilot reported?	Yes
Preference elicitation technique	Willingness-to-pay
Perspective	Own health
End of life definition	"Imminent, premature death from a life threatening disease" (at least six years before respondent's self-reported expected end of life
Life expectancy without treatment attribute levels	Imminent (as above), respondent's self-reported life expectancy
Life expectancy gain from treatment attribute levels	1 QALY worth of life extension (at a quality of life level consistent with respondent's self-reported health)
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No – quality of life was examined but in separate questions
What were respondents choosing between (or choosing to do)?	What the maximum amount they would be willing to pay for a given specific gain

Record	**Pennington *et al.* (2015)**
Was it possible to express indifference?	Yes – respondents could provide the same willingness-to-pay value for multiple gains
Were visual aids used?	Yes
Strength of preference examined at the individual respondent level?	Yes – strength of preference indicated by differing willingness-to-pay amounts
Number of tasks completed by each respondent	5
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Evidence consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	When comparing QALY gains obtained in the near future, life extensions were valued more highly then quality of life improvements; low median values for life extensions at respondents' expected end of life strongly influenced by the large number of observations at zero
Other factors examined	Avoiding quality of life loss now (over 4yrs or 10yrs); avoiding time spent in coma (intended to elicit a gain in longevity occurring in the near future)
Impact of background characteristics reported?	Yes – older age and poorer health associated with lower willingness-to-pay values for life extension in imminent death scenario
Were qualitative data or explanatory factors sought?	No / not reported
Was any reference made to age-related preferences?	Yes – hypothetical scenarios was based on respondents' actual ages and self-reported life expectancies
Was any reference made to time-related preferences?	Yes – authors acknowledge that gains in the future would be discounted, and that for an individual facing immediate death the normal opportunity cost considerations may not apply

Record	**Pinto-Prades *et al.* (2014)**
Year of publication	2014
Year of study conduct	2010
Country or countries of origin of data	Spain
Sample size	813
Type of sample	Public
Sample recruitment process	Door-knock
Criteria for excluding respondents and/or observations reported?	Yes – excluded protest responders in the willingness-to-pay tasks
Mode of administration	Computer-assisted personal interview
Summary of primary study objective(s)	To test for support for end of life prioritisation
Was end of life (or a related term) mentioned explicitly in the study objectives?	Yes
Pilot reported?	Yes
Preference elicitation technique	Willingness-to-pay and person trade-off
Perspective	Both – own health (willingness-to-pay tasks); social decision-maker (person trade-off tasks)
End of life definition	Life expectancy without treatment
Life expectancy without treatment attribute levels	3mths, 6mths, 18mths
Life expectancy gain from treatment attribute levels	6mths, 18mths
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	Yes
What were respondents choosing between (or choosing to do)?	What the maximum amount they would be willing to pay for a 10% chance of improving their condition in a specified way (willingness-to-pay tasks); the number of patients treated of one type they consider equivalent to treating one patient of another type (person trade-off tasks)

Record	**Pinto-Prades *et al.* (2014)**
Was it possible to express indifference?	Yes – respondents could provide the same willingness-to-pay value for multiple gains or choose an equal number of both types of patient in the person trade-off task
Were visual aids used?	Yes
Strength of preference examined at the individual respondent level?	Yes – strength of preference indicated by differing willingness-to-pay amounts and levels of trade-off
Number of tasks completed by each respondent	6
Time taken to complete survey reported?	Yes – 21 minutes on average
Summary of finding: end of life vs. non-end of life	Evidence consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Quality of life improvement preferred
Other results of potential interest	Reasonably large proportion of respondents did not give too much value to a short life extension but those who did were willing to pay quite a lot (similar split of opinion observed in PTO responses)
Other factors examined	None
Impact of background characteristics reported?	No
Were qualitative data or explanatory factors sought?	No / not reported
Was any reference made to age-related preferences?	No
Was any reference made to time-related preferences?	No

Record	**Richardson *et al.* (2012)**
Year of publication	2012
Year of study conduct	2009-2010
Country or countries of origin of data	Australia
Sample size	544
Type of sample	Public
Sample recruitment process	Internet panel; targeted postal invitations (based on socioeconomic characteristics of residential postcodes)
Criteria for excluding respondents and/or observations reported?	Yes – excluded respondents whose comments or answers indicated misunderstanding
Mode of administration	Multiple modes: internet survey; self-completion paper survey (postal)
Summary of primary study objective(s)	To test a technique for measuring support for health-maximisation and health sharing
Was end of life (or a related term) mentioned explicitly in the study objectives?	No
Pilot reported?	No
Preference elicitation technique	Novel cross between a discrete choice and budget allocation exercise
Perspective	Social decision-maker
End of life definition	Immediate death without treatment
Life expectancy without treatment attribute levels	Multiples of 4yrs and 6yrs
Life expectancy gain from treatment attribute levels	4yrs, 6yrs, 8yrs, 12yrs
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No
What were respondents choosing between (or choosing to do)?	Which of four patients to give a life extension to
Was it possible to express indifference?	No

Record	**Richardson *et al.* (2012)**
Were visual aids used?	Yes
Strength of preference examined at the individual respondent level?	Not reported / unclear
Number of tasks completed by each respondent	1 (comprising 18 to 29 iterations)
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Mixed or inconclusive evidence
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	Results indicate that respondents were primarily concerned with outcome egalitarianism (as opposed to maximising health outcomes)
Other factors examined	Sharing / outcome egalitarianism
Impact of background characteristics reported?	Yes – none found to influence propensity to favour treating end of life patients
Were qualitative data or explanatory factors sought?	No / not reported
Was any reference made to age-related preferences?	No – but all patients start at the same age (25yrs), so the results could be interpreted in terms of desire to equalise expected age at death
Was any reference made to time-related preferences?	Yes – authors acknowledge that there may be some variation from the orthodox economic prediction if time discounting is taken into account

Record	**Rowen *et al.* (2016a)**
Year of publication	2015
Year of study conduct	Not reported
Country or countries of origin of data	UK
Sample size	3,669
Type of sample	Public
Sample recruitment process	Internet panel
Criteria for excluding respondents and/or observations reported?	No exclusions
Mode of administration	Internet survey
Summary of primary study objective(s)	To test for support for multiple prioritisation criteria
Was end of life (or a related term) mentioned explicitly in the study objectives?	Yes
Pilot reported?	Yes
Preference elicitation technique	Discrete choice experiment
Perspective	Social decision-maker
End of life definition	Life expectancy without treatment; dummy variable in the modelling representing the cut-offs associated with the NICE criteria
Life expectancy without treatment attribute levels	3mths, 6mths, 9mths, 1yr, 2yrs, 5yrs, 10yrs, 30yrs, 60yrs
Life expectancy gain from treatment attribute levels	0mths, 1mth, 3mths, 6mths, 9mths, 1yr, 3yrs, 10yrs, 60yrs
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No
What were respondents choosing between (or choosing to do)?	Which of two patient groups to treat

Record	**Rowen *et al.* (2016a)**
Was it possible to express indifference?	No
Were visual aids used?	Yes
Strength of preference examined at the individual respondent level?	No
Number of tasks completed by each respondent	10 (+further attitudinal questions)
Time taken to complete survey reported?	Yes – 21 minutes on average
Summary of finding: end of life vs. non-end of life	Evidence consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	Preference for larger QALY gains, but at a diminishing rate; some support for prioritising those with higher burden of illness, though not robust
Other factors examined	QALY gain, burden of illness
Impact of background characteristics reported?	No
Were qualitative data or explanatory factors sought?	Yes – in piloting and via attitudinal questions
Was any reference made to age-related preferences?	No – age attribute was purposely omitted
Was any reference made to time-related preferences?	No

Record	**Rowen *et al.* (2016b)**
Year of publication	2016
Year of study conduct	Not reported
Country or countries of origin of data	UK
Sample size	371
Type of sample	Public
Sample recruitment process	Door-knock; internet panel
Criteria for excluding respondents and/or observations reported?	No exclusions
Mode of administration	Multiple modes: non-computer-assisted personal interview (except in some arms where the introductory video was shown on a computer); internet survey
Summary of primary study objective(s)	To test for framing and mode of administration effects in the elicitation of preferences regarding burden of illness [a]
Was end of life (or a related term) mentioned explicitly in the study objectives?	No
Pilot reported?	No
Preference elicitation technique	Other choice exercise
Perspective	Social decision-maker
End of life definition	Life expectancy without treatment / due to condition
Life expectancy without treatment attribute levels	5yrs, 10yrs, 15yrs (but the sole end of life vs. non-end of life question involved a choice between patients with life expectancies of 5yrs and 10yrs, respectively)
Life expectancy gain from treatment attribute levels	0yrs, 1yr, 2yrs (but in the sole end of life vs. non-end of life question, life expectancy gain was 1yr for both candidate recipient groups)
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	Yes
What were respondents choosing between (or choosing to do)?	Which of two patient groups to treat

Record	**Rowen *et al.* (2016b)**
Was it possible to express indifference?	No
Were visual aids used?	Yes (in four of six arms; n=240); no (in two of six arms; n=131)
Strength of preference examined at the individual respondent level?	No
Number of tasks completed by each respondent	9 (2 of which were practice tasks, but were reported in full by authors)
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Evidence not consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported (two questions involved choices between quality of life improvements and life extensions, but life expectancy without treatment was set to 10/15 years so is deemed not to describe an end of life context)
Other results of potential interest	Responses were affected by mode of administration but not by question wording or use of visual aids
Other factors examined	QALY gain (size and type); burden of illness
Impact of background characteristics reported?	No – not for question of relevance to end of life (for other questions, few sociodemographic variables were significant)
Were qualitative data or explanatory factors sought?	No – follow-up questions were asked but these focused on framing issues and task understanding rather than on reasons for choices
Was any reference made to age-related preferences?	No
Was any reference made to time-related preferences?	No

[a] The only task in this study relevant to the research question underpinning the literature review was labelled as a 'practice question'. However, in the paper the authors do not treat the practice question as any less valid or reliable than the main (non-practice) questions, and present a full analysis of the responses to the practice questions. A notable feature of the practice questions in this study was that respondents were, in effect, asked to reconsider and confirm their responses. This suggests that the responses should not be interpreted as constituting lower quality data than the responses to the main questions. The decision to include this study in the review was informed by a discussion with one of the study authors (Tsuchiya, A., 2016, personal communication, 20 September).

Record	**Shah *et al.* (2014)**
Year of publication	2014
Year of study conduct	2011
Country or countries of origin of data	UK
Sample size	50
Type of sample	Public
Sample recruitment process	Door-knock
Criteria for excluding respondents and/or observations reported?	No
Mode of administration	Non-computer-assisted personal interview
Summary of primary study objective(s)	To test for support for end of life prioritisation
Was end of life (or a related term) mentioned explicitly in the study objectives?	Yes
Pilot reported?	Yes
Preference elicitation technique	Other choice exercise
Perspective	Social decision-maker
End of life definition	Life expectancy without treatment
Life expectancy without treatment attribute levels	1yr, 10yrs
Life expectancy gain from treatment attribute levels	6mths, 1yr
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	Yes
What were respondents choosing between (or choosing to do)?	Which of two patients to treat
Was it possible to express indifference?	Yes – an 'I have no preference' option was available
Were visual aids used?	Yes

Record	**Shah *et al.* (2014)**
Strength of preference examined at the individual respondent level?	No
Number of tasks completed by each respondent	6
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Evidence consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Quality of life improvement preferred
Other results of potential interest	No evidence that age- or time-related preferences are motivating factors for choosing to treat end of life patient; no evidence that concern about the life stage of end of life patients is a motivating factor for preferring either life-extending or quality of life-improving treatments for those patients
Other factors examined	Age, time preference
Impact of background characteristics reported?	No
Were qualitative data or explanatory factors sought?	Yes – respondents indicated the reasons for their choices by choosing from a list
Was any reference made to age-related preferences?	Yes – no evidence that concern about age is a motivating factor for choosing to treat end of life patient
Was any reference made to time-related preferences?	Yes – one task involved choosing between a patient who had known their prognosis for some time and another who had only just learned their prognosis (life expectancy without treatment was the same for both)

Record	**Shah *et al.* (2015a)**
Year of publication	2015
Year of study conduct	2012
Country or countries of origin of data	UK
Sample size	3,969
Type of sample	Public
Sample recruitment process	Internet panel
Criteria for excluding respondents and/or observations reported?	Yes – excluded respondents who spent insufficient time completing the survey
Mode of administration	Internet survey
Summary of primary study objective(s)	To test for support for end of life prioritisation
Was end of life (or a related term) mentioned explicitly in the study objectives?	Yes
Pilot reported?	Yes
Preference elicitation technique	Discrete choice experiment
Perspective	Social decision-maker
End of life definition	Life expectancy without treatment; dummy variable in the modelling representing the cut-offs associated with the NICE criteria
Life expectancy without treatment attribute levels	3mths, 12mths, 24mths, 36mths, 60mths
Life expectancy gain from treatment attribute levels	0mths, 1mth, 2mths, 3mths, 6mths, 12mths
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	Yes
What were respondents choosing between (or choosing to do)?	Which of two patients to treat

Record	**Shah *et al.* (2015a)**
Was it possible to express indifference?	No
Were visual aids used?	Yes
Strength of preference examined at the individual respondent level?	No
Number of tasks completed by each respondent	10 (+2 further tasks examining the issue of preparedness)
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Evidence not consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Life extension preferred
Other results of potential interest	Majority of respondents supported a mixture of the QALY-maximisation and priority-to-worst-off approaches to priority-setting
Other factors examined	Quality of life without treatment, preparedness
Impact of background characteristics reported?	Yes – no characteristics found to be associated with preferences
Were qualitative data or explanatory factors sought?	No / not reported
Was any reference made to age-related preferences?	No – age attribute was purposely omitted
Was any reference made to time-related preferences?	'Time with knowledge' attribute was examined but the results were not reported; authors note that applying a positive discount rate would likely further strengthen their finding of a lack of support for an end of life premium

Record	**Shiroiwa *et al.* (2010)**
Year of publication	2010
Year of study conduct	2007-2008
Country or countries of origin of data	Australia, Japan, Korea, Taiwan, UK, USA
Sample size	5,620
Type of sample	Public
Sample recruitment process	Internet panel
Criteria for excluding respondents and/or observations reported?	Yes – recruited respondents aged 20 to 59 years only (thereby excluding individuals aged 60 years and older)
Mode of administration	Internet survey
Summary of primary study objective(s)	To obtain the monetary value of a QALY (in six countries)
Was end of life (or a related term) mentioned explicitly in the study objectives?	No
Pilot reported?	Yes
Preference elicitation technique	Willingness-to-pay
Perspective	Both – own health (end of life and non-end of life scenarios); social decision-maker (end of life scenario only)
End of life definition	Serious illness that immediately threatens [your / their] life
Life expectancy without treatment attribute levels	0yrs, 5yrs
Life expectancy gain from treatment attribute levels	1yr
Was disease labelled or named?	No (but disease was described as a life-limiting illness such as metastatic cancer)
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No
What were respondents choosing between (or choosing to do)?	Whether or not to pay set amounts for a given life extension
Was it possible to express indifference?	Yes – respondents could provide the same willingness-to-pay value for multiple gains

Record	**Shiroiwa *et al.* (2010)**
Were visual aids used?	No
Strength of preference examined at the individual respondent level?	Yes – strength of preference indicated by differing willingness-to-pay amounts
Number of tasks completed by each respondent	4
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Evidence consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	In Japan and Korea, the highest willingness-to-pay values observed were for a life extension for a family member; in Australia, UK and USA, the highest willingness-to-pay values observed were for a life extension for an unidentified member of society
Other factors examined	Willingness-to-pay for a life extension for a family member and for an unidentified member of society facing life-threatening illness
Impact of background characteristics reported?	Yes – respondents with high household income and education levels gave higher willingness-to-pay values for life extensions at the end of life
Were qualitative data or explanatory factors sought?	No / not reported
Was any reference made to age-related preferences?	No
Was any reference made to time-related preferences?	Yes – authors interpret difference between willingness-to-pay values in end of life and non-end of life scenarios in terms of time preference, and use the data to estimate discount rates for each country (ranging from 1.6% to 6.8%)

Record	**Shiroiwa *et al.* (2013)**
Year of publication	2013
Year of study conduct	2011
Country or countries of origin of data	Japan
Sample size	2,283
Type of sample	Public
Sample recruitment process	Internet panel
Criteria for excluding respondents and/or observations reported?	Yes – recruited respondents aged 20 to 69 years only (thereby excluding individuals aged 70 years and older)
Mode of administration	Internet survey
Summary of primary study objective(s)	To obtain the monetary value of a QALY
Was end of life (or a related term) mentioned explicitly in the study objectives?	No
Pilot reported?	No
Preference elicitation technique	Willingness-to-pay
Perspective	Own health
End of life definition	Life expectancy without treatment (end of life scenario 1); life-threatening situation (end of life scenario 2)
Life expectancy without treatment attribute levels	1mth
Life expectancy gain from treatment attribute levels	2mths, 4mths, 7mths, 14mths
Was disease labelled or named?	No
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No – questions examining quality of life improvement were included, but these were related to non-end of life scenarios
What were respondents choosing between (or choosing to do)?	Whether or not to pay set amounts for a given specific gain
Was it possible to express indifference?	Yes – respondents could provide the same willingness-to-pay value for multiple gains

Record	**Shiroiwa *et al.* (2013)**
Were visual aids used?	No
Strength of preference examined at the individual respondent level?	Yes – strength of preference indicated by differing willingness-to-pay amounts
Number of tasks completed by each respondent	1
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Evidence not consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	Monetary value of a QALY is higher for severe health states than for mild health states
Other factors examined	Avoiding quality of life loss now (for periods lasting between 4 and 20 months)
Impact of background characteristics reported?	No – not specifically for questions of relevance to end of life (overall, willingness-to-pay values were significantly correlated with household income)
Were qualitative data or explanatory factors sought?	No / not reported
Was any reference made to age-related preferences?	No
Was any reference made to time-related preferences?	No

Record	**Skedgel *et al.* (2014)**
Year of publication	2015
Year of study conduct	2011-2012
Country or countries of origin of data	Canada
Sample size	656
Type of sample	Public, decision-makers
Sample recruitment process	Internet panel (public); flyers and email invitations (decision-makers)
Criteria for excluding respondents and/or observations reported?	No exclusions
Mode of administration	Internet survey
Summary of primary study objective(s)	To test for support for multiple prioritisation criteria
Was end of life (or a related term) mentioned explicitly in the study objectives?	No
Pilot reported?	Yes
Preference elicitation technique	Discrete choice experiment
Perspective	Social decision-maker
End of life definition	Life expectancy without treatment
Life expectancy without treatment attribute levels	1mth, 5yrs, 10yrs
Life expectancy gain from treatment attribute levels	1yr, 5yrs, 10yrs
Was disease labelled or named?	Yes – cancer
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No
What were respondents choosing between (or choosing to do)?	Which of two health programmes to allocate (all of) a fixed budget to
Was it possible to express indifference?	No
Were visual aids used?	Yes

Record	**Skedgel *et al.* (2014)**
Strength of preference examined at the individual respondent level?	No
Number of tasks completed by each respondent	11
Time taken to complete survey reported?	Yes – 9.5 minutes on average (public)
Summary of finding: end of life vs. non-end of life	Evidence not consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	Evidence of support for treating younger and larger patient groups; and for deprioritising treatment for those who will be in poor health after treatment
Other factors examined	Age, quality of life without treatment, quality of life with treatment, number of patients treated
Impact of background characteristics reported?	Yes – background characteristics were not statistically significantly associated with (latent) class membership
Were qualitative data or explanatory factors sought?	No / not reported
Was any reference made to age-related preferences?	Yes – evidence of support for treating younger patients, though the author did not interact the age and life expectancy without treatment variables
Was any reference made to time-related preferences?	No

Record	**Stahl *et al.* (2008)**
Year of publication	2008
Year of study conduct	Not reported
Country or countries of origin of data	USA
Sample size	623
Type of sample	Public
Sample recruitment process	Internet panel
Criteria for excluding respondents and/or observations reported?	No
Mode of administration	Internet survey
Summary of primary study objective(s)	To test for support for multiple prioritisation criteria
Was end of life (or a related term) mentioned explicitly in the study objectives?	Yes
Pilot reported?	No
Preference elicitation technique	Other choice exercise
Perspective	Social decision-maker
End of life definition	Urgency (life expectancy without treatment)
Life expectancy without treatment attribute levels	Levels not reported explicitly, but appear to cover: <1mth, 3mths, 6mths, 9mths, 12mths, 15mths, 18mths, 21mths, 24mths
Life expectancy gain from treatment attribute levels	1yr, 2yrs, 3yrs, 4yrs, 5yrs, 6yrs, 7yrs, 8yrs, 9yrs, 10yrs
Was disease labelled or named?	Yes – organ transplantation
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No
What were respondents choosing between (or choosing to do)?	Which of two patients to give an organ transplant to
Was it possible to express indifference?	Yes – a 'can't decide' option was available

Record	**Stahl *et al.* (2008)**
Were visual aids used?	No
Strength of preference examined at the individual respondent level?	Yes – attribute levels were varied incrementally
Number of tasks completed by each respondent	33 (unclear whether each respondent answered all or a subset of the 33)
Time taken to complete survey reported?	No
Summary of finding: end of life vs. non-end of life	Evidence consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	When both patients have better (worse) than average quality of life, respondents preferred to treat the worse off (better off) patient
Other factors examined	Age, life expectancy with treatment, quality of life without treatment, quality of life with treatment; single-factor and cross-factor trade-offs examined
Impact of background characteristics reported?	Yes – older (>40yrs) and female respondents had narrower windows of indifference – i.e. preferred to treat end of life patient until the difference between the life expectancies of the patients was extremely small; respondents with transplant recipient in family placed greater importance on quality of life without treatment then life expectancy without treatment unless the latter was extremely short (<1mth)
Were qualitative data or explanatory factors sought?	No / not reported
Was any reference made to age-related preferences?	Yes – for an older patient to receive priority over a younger patient, the older patient must be at least 2.5mths closer to their end of life than the younger patient
Was any reference made to time-related preferences?	No

Record	**Stolk *et al.* (2005)**
Year of publication	2005
Year of study conduct	Not reported
Country or countries of origin of data	Netherlands
Sample size	65
Type of sample	Students, researchers, health policy makers
Sample recruitment process	Not reported
Criteria for excluding respondents and/or observations reported?	No
Mode of administration	Non-computer-assisted personal interview
Summary of primary study objective(s)	To test for support for multiple prioritisation criteria
Was end of life (or a related term) mentioned explicitly in the study objectives?	No
Pilot reported?	Yes
Preference elicitation technique	Other choice exercise
Perspective	Social decision-maker
End of life definition	Information on life expectancy without treatment not provided explicitly but could be calculated given information on age, life expectancy (disease-free and with disease) and life years lost due to disease
Life expectancy without treatment attribute levels	0.5yrs, 2.25yrs, 3yrs, 11yrs, 14yrs, 14.5yrs, 16yrs, 20yrs, 20.5yrs, 22.5yrs
Life expectancy gain from treatment attribute levels	N/A – treated patient would be given a 'wonder pill' which would relieve them of all described health problems and bring them back to normal health
Was disease labelled or named?	Yes – each patient had a different disease
Did the study examine whether quality of life improving or life extending treatments are preferred for end of life patients?	No
What were respondents choosing between (or choosing to do)?	Which of two patients to treat

Record	**Stolk *et al.* (2005)**
Was it possible to express indifference?	No
Were visual aids used?	No
Strength of preference examined at the individual respondent level?	No
Number of tasks completed by each respondent	45
Time taken to complete survey reported?	Yes – 20 minutes on average
Summary of finding: end of life vs. non-end of life	Evidence not consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Not examined / reported
Other results of potential interest	Fair innings and (to a lesser extent) proportional shortfall approaches to priority-setting were highly correlated with the observed rank order implied by respondents' choices
Other factors examined	Fair innings, severity, proportional shortfall
Impact of background characteristics reported?	Yes – there were no major differences in the rank orderings of the three respondent subgroups
Were qualitative data or explanatory factors sought?	No / not reported
Was any reference made to age-related preferences?	Yes – treatments for elderly patients were not valued as higher prospective health theories that ignore the past (i.e. age) would have predicted
Was any reference made to time-related preferences?	No

Appendix 4 Empirical study 1 survey instrument

Health care priority setting preference project

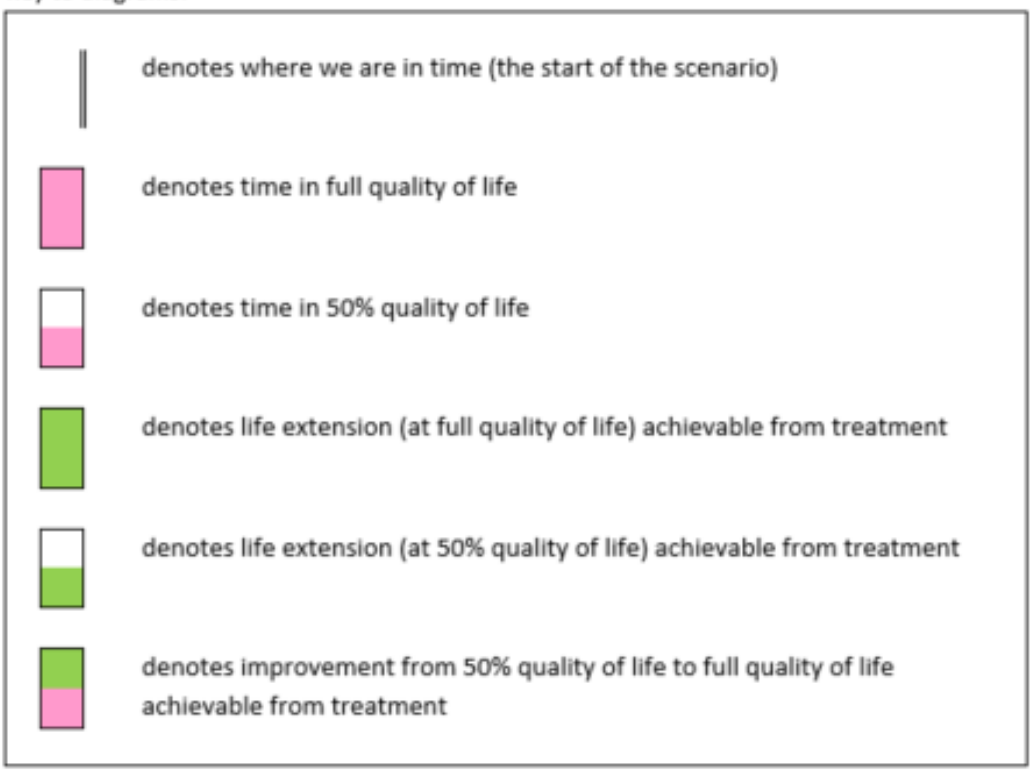
ID: _ _ _ _

Date: _ _ / _ _ / _ _

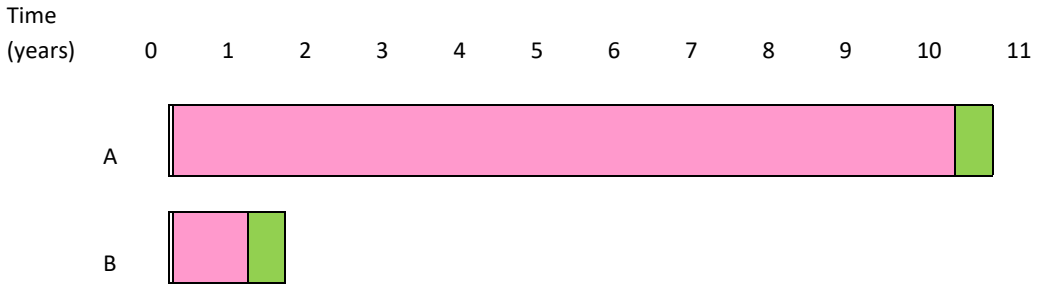
Time: _ _ : _ _

There are five scenarios in this questionnaire. Please read the description for each scenario carefully. Please let the interviewer know if you have any questions at any stage of the interview. Remember, there are no right or wrong answers – we are simply seeking your views.

Key to diagrams:



Scenario 1



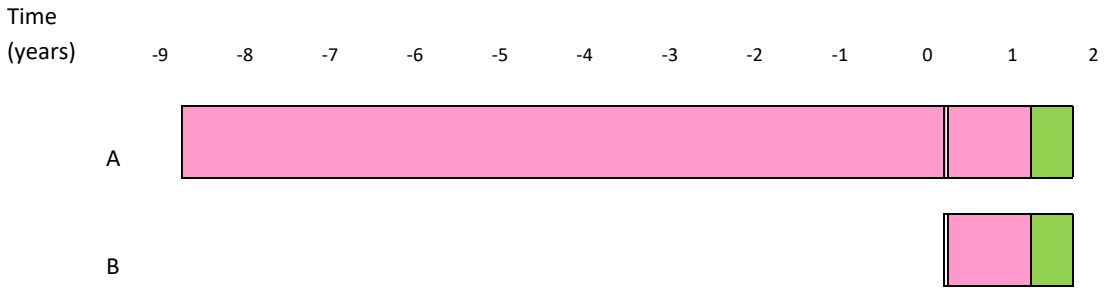
Consider two patients, patient A and patient B, who are the same age as each other. Suppose that both patient A and patient B have just been diagnosed with illnesses. The illnesses are asymptomatic – that is, they have no effect on the patient's quality of life.

Patient A will live for 10 years, from today, before dying. Patient B will live for 1 year, from today, before dying.

There is a treatment, which, if taken today, would extend the life of either patient A or patient B by 6 months. Treatment would not affect either patient's quality of life. However, the health service has only enough funds to treat one of the two patients, and there are no alternative treatments available (furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today).

Would you prefer to treat patient A or patient B?

Scenario 2



Consider two patients, patient A and patient B, who are the same age as each other. Patient B has just been diagnosed with an illness; patient A has an illness which he/she was diagnosed with 9 years ago. Both illnesses are asymptomatic – that is, they have no effect on the patient's quality of life.

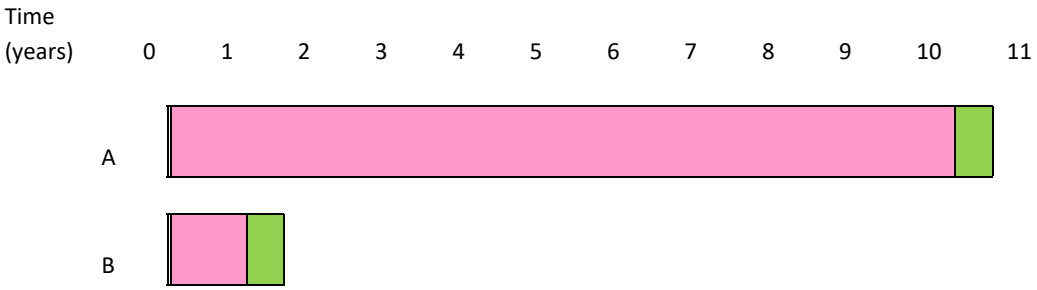
Patient A was told 9 years ago that he/she will live for 10 years before dying. This means that from today, he/she will live for 1 year before dying.

Patient B has been told that he/she will live for 1 year, from today, before dying.

There is a treatment, which, if taken today, would extend the life of either patient A or patient B by 6 months. Treatment would not affect either patient's quality of life. However, the health service has only enough funds to treat one of the two patients, and there are no alternative treatments available (furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today).

Would you prefer to treat patient A or patient B?

Scenario 3



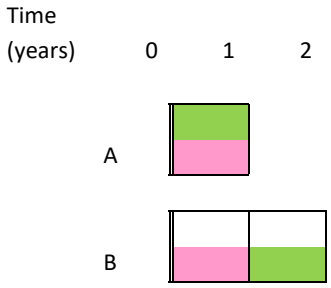
Consider two patients, patient A and patient B. Suppose that both patient A and patient B have just been diagnosed with illnesses. The illnesses are asymptomatic – that is, they have no effect on the patient's quality of life.

Patient A will live for 10 years, from today, before dying. Patient B will live for 1 year, from today, before dying. Patient B is 9 years older than patient A, so both patients will die at the same age without treatment.

There is a treatment, which, if taken today, would extend the life of either patient A or patient B by 6 months. Treatment would not affect either patient's quality of life. However, the health service has only enough funds to treat one of the two patients, and there are no alternative treatments available (furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today).

Would you prefer to treat patient A or patient B?

Scenario 4



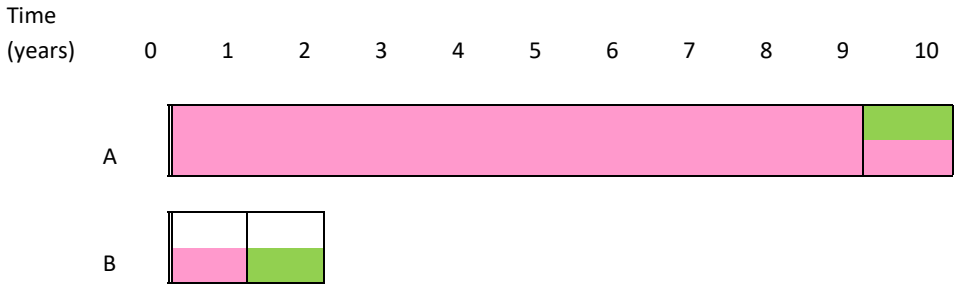
Consider two patients, patient A and patient B, who are the same age as each other. Suppose that both patient A and patient B have just been diagnosed with illnesses.

Patient A will live for 1 year, from today, before dying. Patient B will also live for 1 year, from today, before dying. The illnesses have a negative impact on quality of life – both patients will experience their final year of life at 50% of full health.

There is a treatment, which, if taken today, would restore patient A to full health. It would not affect patient A's life expectancy. Another treatment would, if taken today, extend the life of patient B by 1 year. It would not affect patient B's quality of life, so patient B's remaining life would be lived at 50% health. The health service has only enough funds to treat one of the two patients, and there are no alternative treatments available (furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today).

Would you prefer to treat patient A or patient B?

Scenario 5



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 10 years, from today, before dying. Patient B will live for 1 year, from today, before dying. The illnesses have a negative impact on quality of life – both patients will experience their final year of life at 50% of full health. Patient B is 9 years older than patient A, so both patients will die at the same age without treatment.

There is a treatment, which, if taken today, would restore patient A to full health in her final year of life. It would not affect patient A's life expectancy. The same treatment would, if taken today, extend the life of patient B by 1 year. It would not affect patient B's quality of life, so patient B's remaining life would be lived at 50% health.

The health service has only enough funds to treat one of the two patients, and there are no alternative treatments available (furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today).

Would you prefer to treat patient A or patient B?

Appendix 5 Empirical study 1 information sheet

1. Research Project Title:

Health care priority setting preference project

2. Invitation paragraph

You are being invited to take part in a research project. Before you decide whether you wish to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Take time to decide whether or not you wish to take part. Thank you for reading this.

3. What is the project's purpose?

The purpose of the project is to find out what the general public thinks about a range of hypothetical scenarios where health care decision makers have to choose which types of treatment to allocate funding to. Better understanding of public preferences will help organisations such as the National Institute for Health and Clinical Excellence (NICE) to make appropriate resource allocation decisions.

4. Why have I been chosen?

We are seeking to survey around 20 students and staff at the University of Sheffield as a pilot study.

5. Do I have to take part?

It is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep (and be asked to sign a consent form) and you can still withdraw at any time without it affecting any benefits that you are entitled to in any way. If you decide to stop, then any information that you have provided will be destroyed. You do not have to give a reason for not taking part.

6**. What will happen to me if I take part?**

If you agree to take part, you will complete an interview survey that should last for no more than 30 minutes. You will be interviewed by a researcher, who will ask you a series of questions which require you to compare hypothetical scenarios in which a health care decision maker must allocate resources to one of two treatments for ill health. A paper questionnaire will be used to provide illustrations of the scenarios and to record your responses. You will also be asked some questions about your thoughts and opinions about the exercise, as well as some questions about your experience of ill health.

You will only be asked to participate in one interview.

7. What do I have to do?

You will be asked to answer a series of questions involving hypothetical scenarios. There are no right or wrong answers – we are simply seeking your views.

8. What are the possible disadvantages and risks of taking part?

Some participants may feel uncomfortable when asked to think about scenarios involving illness and death. However, previous research in this area has shown that participants are generally interested and engaged when taking part in these types of exercises.

9. What are the possible benefits of taking part?

All participants will be given a small cash payment for agreeing to take part in the study. You will also be contributing to research that will help health care decision makers to better understand the preferences of members of the general public.

10. What if something goes wrong?

Should you wish to raise a complaint regarding your treatment by researchers or any other aspect of the study, you should contact Professor Aki Tsuchiya (a.tsuchiya@sheffield.ac.uk), who will follow up on your complaint immediately. However, should you feel that your complaint has not been handled to your satisfaction, you can contact the University's Registrar and Secretary.

11. Will my taking part in this project be kept confidential?

All information which is collected about you during the course of the research will be kept strictly confidential. All survey responses will be anonymised, so you will not be identified in any reports or publications.

12. What will happen to the results of the research project?

The results of the project will be written up in a report for the National Institute for Health and Clinical Excellence (NICE), and will be published in academic journals and presented at conferences. You will not be identified in any reports or publications. The anonymised data collected during the course of the project may be used for additional or subsequent research and analysis.

13. Who is organising and funding the research?

The project is being organised by Allan Wailoo, Aki Tsuchiya, and Koonal Shah, of the University of Sheffield's' School of Health and Related Research. It has been funded by the National Institute for Health and Clinical Excellence.

14. Who has ethically reviewed the project?

The project has been reviewed by the School of Health and Related Research Ethics Committee at the University of Sheffield.

15. Contact for further information

For information regarding participant recruitment and co-ordination of the interview programme, please contact Koonal Shah (k.k.shah@sheffield.ac.uk; 07920 496832). For any other issues, please contact Professor Aki Tsuchiya (a.tsuchiya@sheffield.ac.uk; 0114 222 0710).

16. Will I be recorded, and how will the recorded media be used?

The interviews will be audio recorded. The recordings will be used only for analysis and will be kept in secure premises. No other use will be made of them without your written permission, and no individual outside the project will be allowed access to them. Once the pilot study is written up, the recording will be destroyed.

Appendix 6 Empirical study 2 survey instrument

Interviewer no: | | | | | **Interviewer name:** |

Date: | | / | | **Time interview started:** | | : | | **U** | | |

Main Questionnaire

HAND PARTICIPANT BOOKLET TO RESPONDENT

Thank you for agreeing to take part in this research project.

The interview consists of seven scenarios. I will read the description for each scenario to you, and will then ask you to answer some questions about that scenario. Please listen carefully to the descriptions. You may also refer to the summary tables and diagrams for each scenario, which can be found in the booklet I have given you.

Please let me know if you have any questions at any stage of the interview. Remember, there are no right or wrong answers – we are simply seeking your view.

Consider two patients, patient A and patient B. Both patients are adults, and are the same age as each other. Suppose that both patient A and patient B have just been diagnosed with illnesses. The illnesses are asymptomatic – that is, they have no effect on the patient's health-related quality of life.

Patient A will live for 10 years, from today, before dying. Patient B will live for 1 year, from today, before dying. These life expectancies are shown by the areas shaded pink in the diagrams.

There is a treatment, which, if taken today, would extend the life of patient A by 2 years. The same treatment would extend the life of patient B by 6 months. These gains are shown by the areas shaded green in the diagrams. Treatment would not affect either patient's health-related quality of life. However, the health service has only enough funds to treat one of the two patients, and there are no alternative treatments available. Furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today – this is the only opportunity for treatment.

Please complete the questions for Scenario 1, which can be found on page 3 of your booklet.

Consider two patients, patient A and patient B. Both patients are adults, and are the same age as each other. Suppose that both patient A and patient B have just been diagnosed with illnesses. The illnesses are asymptomatic – that is, they have no effect on the patient's health-related quality of life.

Patient A will live for 10 years, from today, before dying. Patient B will live for 1 year, from today, before dying.

There is a treatment, which, if taken today, would extend the life of either patient A or patient B by 6 months. Treatment would not affect either patient's health-related quality of life. However, the health service has only enough funds to treat one of the two patients, and there are no alternative treatments available. Furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today – this is the only opportunity for treatment.

Please complete the questions for Scenario 2, which can be found on page 5 of your booklet.

Consider two patients, patient A and patient B. Both patients are adults, but patient B is 9 years older than patient A. Suppose that both patient A and patient B have just been diagnosed with illnesses. The illnesses are asymptomatic – that is, they have no effect on the patient's health-related quality of life.

Patient A will live for 10 years, from today, before dying. Patient B will live for 1 year, from today, before dying. Since patient B is 9 years older than patient A, both patients will die at the same age without treatment.

There is a treatment, which, if taken today, would extend the life of either patient A or patient B by 6 months. Treatment would not affect either patient's health-related quality of life. However, the health service has only enough funds to treat one of the two patients, and there are no alternative treatments available. Furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today – this is the only opportunity for treatment.

Please complete the questions for Scenario 3, which can be found on page 7 of your booklet.

Consider two patients, patient A and patient B, who are the same age as each other. Patient B has just been diagnosed with an illness; patient A has an illness which he or she was diagnosed with 9 years ago. Both illnesses are asymptomatic – that is, they have no effect on the patient's health-related quality of life.

Patient A was told 9 years ago that he or she would live for 10 years before dying. This means that from today, he or she will live for 1 year before dying. Patient B has been told that he or she will live for 1 year, from today, before dying.

There is a treatment, which, if taken today, would extend the life of either patient A or patient B by 6 months. Treatment would not affect either patient's health-related quality of life. However, the health service has only enough funds to treat one of the two patients, and there are no alternative treatments available. Furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today – this is the only opportunity for treatment.

Please complete the questions for Scenario 4, which can be found on page 9 of your booklet.

Consider two patients, patient A and patient B, who are both 30 years old. 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 also live for 1 year, from today, before dying. The illnesses have a negative impact on health-related quality of life – both patients will experience their final year of life in a state of ill health. Patients have told us that being in this health state for two years is equally desirable as being in full health for one year – we will therefore call this 50% health.

There is a treatment, which, if taken today, would restore patient A to full health. It would not affect patient A's life expectancy. Another treatment would, if taken today, extend the life of patient B by 1 year. It would not affect patient B's health-related quality of life, so patient B's remaining life would be lived at 50% health.

The health service has only enough funds to treat one of the two patients, and there are no alternative treatments available. Furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today – this is the only opportunity for treatment.

Please complete the questions for Scenario 5, which can be found on page 11 of your booklet.

Consider two patients, patient A and patient B, who are both 70 years old. 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 also live for 1 year, from today, before dying. The illnesses have a negative impact on health-related quality of life – both patients will experience their final year of life in a state of ill health. Patients have told us that being in this health state for two years is equally desirable as being in full health for one year – we will therefore call this 50% health.

There is a treatment, which, if taken today, would restore patient A to full health. It would not affect patient A's life expectancy. Another treatment would, if taken today, extend the life of patient B by 1 year. It would not affect patient B's health-related quality of life, so patient B's remaining life would be lived at 50% health.

The health service has only enough funds to treat one of the two patients, and there are no alternative treatments available. Furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today – this is the only opportunity for treatment.

Please complete the questions for Scenario 6, which can be found on page 13 of your booklet.

Consider two patients, patient A and patient B. Both patients are adults, but patient B is 9 years older than patient A. Suppose that both patient A and patient B have just been diagnosed with illnesses.

Patient A will live for 10 years, from today, before dying. Patient B will live for 1 year, from today, before dying. The illnesses have a negative impact on health-related quality of life – both patients will experience their final year of life in a state of ill health. Patients have told us that being in this health state for two years is equally desirable as being in full health for one year – we will therefore call this 50% health. Since patient B is 9 years older than patient A, both patients will die at the same age without treatment.

There is a treatment, which, if taken today, would restore patient A to full health in his or her final year of life. It would not affect patient A's life expectancy. The same treatment would, if taken today, extend the life of patient B by 1 year. It would not affect patient B's health-related quality of life, so patient B's remaining life would be lived at 50% health.

The health service has only enough funds to treat one of the two patients, and there are no alternative treatments available. Furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today – this is the only opportunity for treatment.

Please complete the questions for Scenario 7, which can be found on page 15 of your booklet.

Thank you for your help in this research

This research was conducted under the terms of the MRS code of conduct and is completely confidential. If you would like to confirm my credentials or those of Accent please call the MRS free on 0500 396999. **HAND OVER THE THANK YOU SLIP AND CASH INCENTIVE.**

Please can I confirm your name and where we can contact you for quality control purposes?

Respondent name:

Telephone: home: work:

RECORD PARTICIPANT URN ON PARTICIPANT BOOKLET

Thank you

INTERVIEWER: PLEASE FILL IN THE FOLLOWING (DO NOT ASK OF OR READ TO RESPONDENTS)

How well do you think the respondent understood and carried out the tasks during the interview?

- Understood and performed tasks easily
- Some problems but seemed to understand the tasks in the end
- Doubtful whether the respondent understood the tasks

In terms of effort and concentration, which one of the following statements best describes the way the respondent undertook the tasks?

- Concentrated very hard and put a great deal of effort into it
- Concentrated fairly hard and put some effort into it
- Didn't concentrate very hard and put little effort into it
- Concentrated at the beginning but lost interest/concentration before reaching the end

Which of the following statements below best describes the environment in which the interview was conducted?

- Quiet and no distraction
- No interruption but some background distraction
- Disruptions and interruptions

I confirm that this interview was conducted under the terms of the MRS code of conduct and is completely confidential

Interviewer's signature:

Time Interview completed: | | | | | |

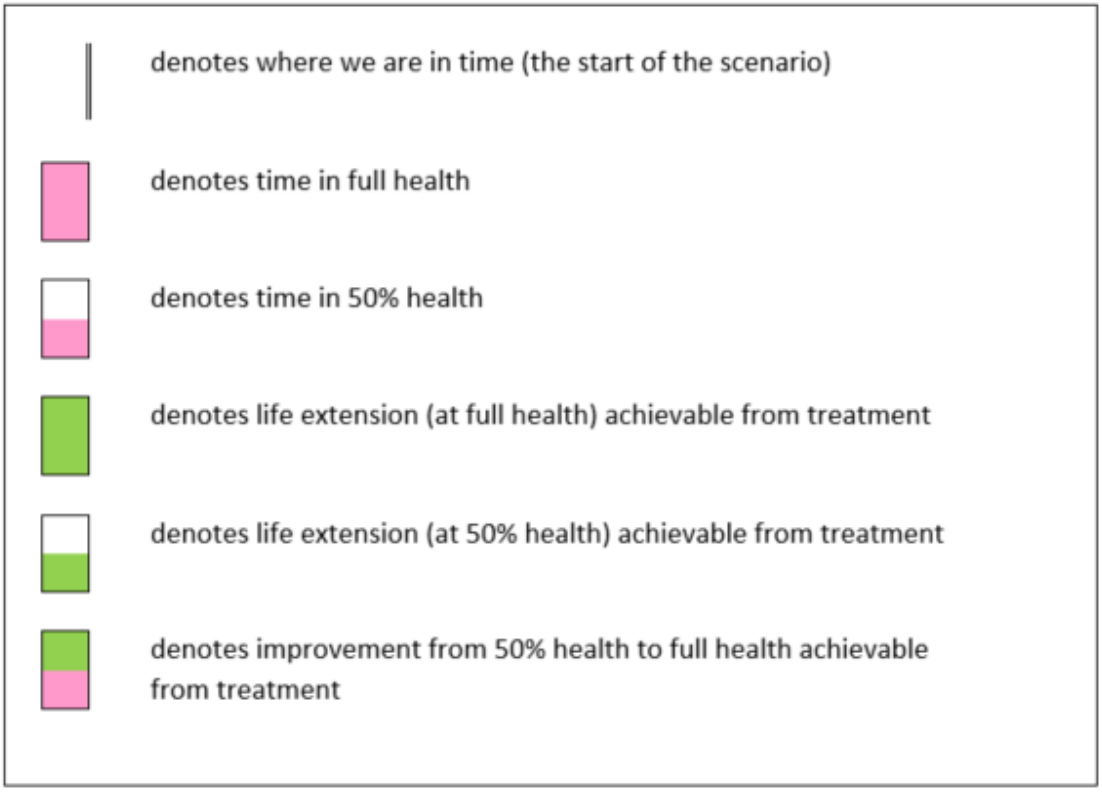
Health care priority setting preference project

Participant booklet

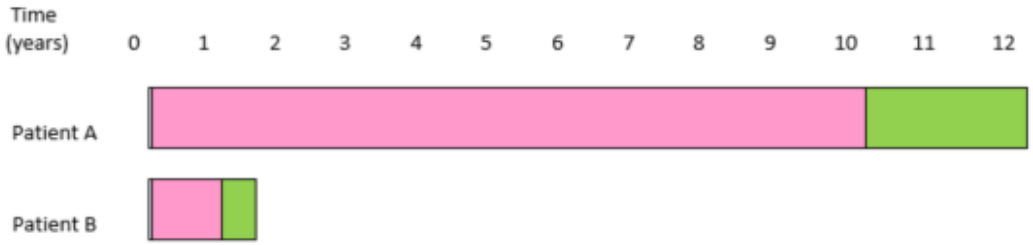
Box to be completed by the interviewer

Participant ID:
Interviewer ID:
Date:
Time:

Key to diagrams

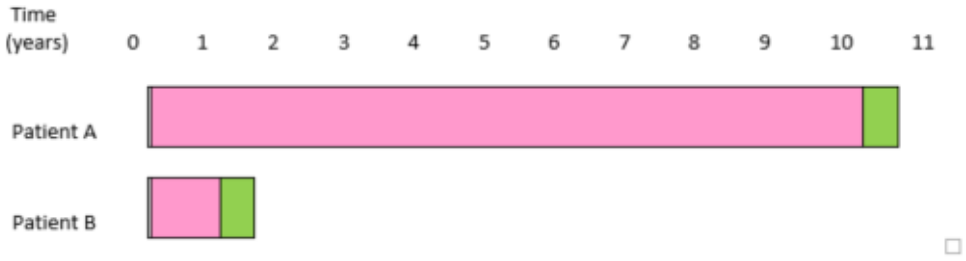


Scenario 0



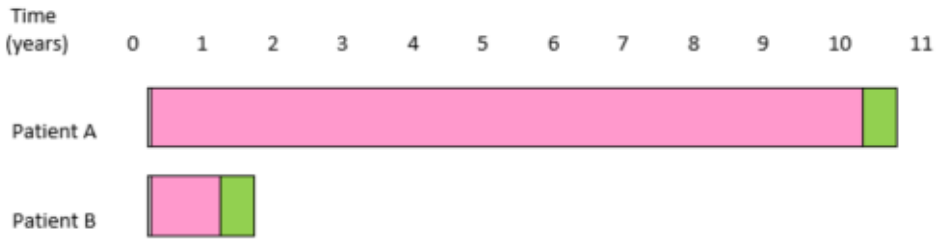
	Patient A	Patient B
Age today	Both patients are same age (adults)	
Age at death without treatment	Patient A will be 9 years older than Patient B	
Life expectancy without treatment (from today)	10 years	1 year
Gain from treatment	+ 2 years	+ 6 months

Scenario 1



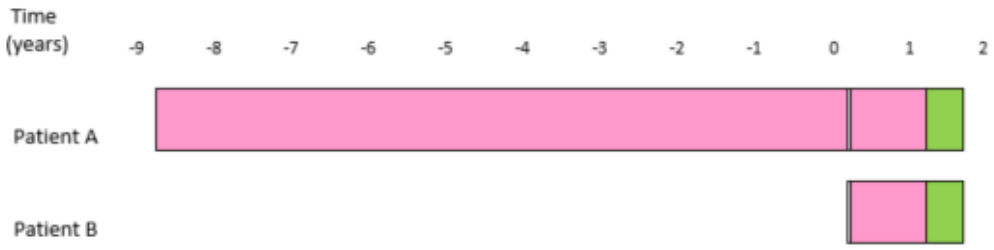
	Patient A	Patient B
Age today	Both patients are same age (adults)	
Age at death without treatment	Patient A will be 9 years older than Patient B	
Life expectancy without treatment (from today)	10 years	1 year
Gain from treatment	+ 6 months	+ 6 months

Scenario 2



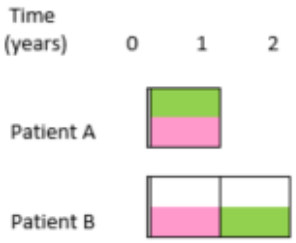
	Patient A	Patient B
Age today	Patient B is 9 years older than Patient A (both adults)	
Age at death without treatment	Both patients will die at the same age	
Life expectancy without treatment (from today)	10 years	1 year
Gain from treatment	+ 6 months	+ 6 months

Scenario 3



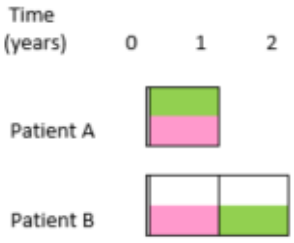
	Patient A	Patient B
Age today	Both patients are same age (adults)	
Age at death without treatment	Both patients will die at the same age	
Timing of diagnosis	9 years ago	Today
Life expectancy without treatment (from today)	1 year	1 year
Gain from treatment	+ 6 months	+ 6 months

Scenario 4



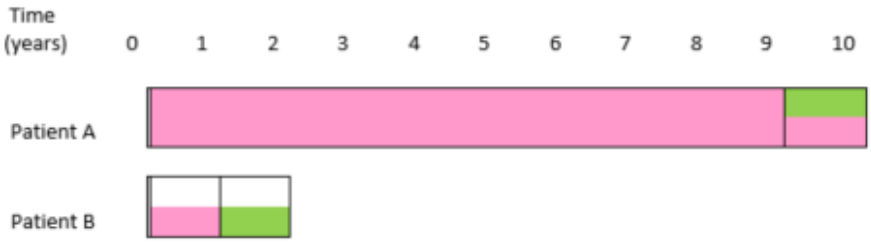
	Patient A	Patient B
Age today	Both patients are the same age (30 years)	
Age at death without treatment	Both patients will die at the same age (31 years)	
Life expectancy without treatment (from today)	1 year	1 year
Health without treatment	50%	50%
Gain from treatment	No life extension Restore to 100% health	+ 1 year No health improvement

Scenario 5



	Patient A	Patient B
Age today	Both patients are the same age (70 years)	
Age at death without treatment	Both patients will die at the same age (71 years)	
Life expectancy without treatment (from today)	1 year	1 year
Health without treatment	50%	50%
Gain from treatment	No life extension Restore to 100% health	+ 1 year No health improvement

Scenario 6



	Patient A	Patient B
Age today	Patient B is 9 years older than Patient A (both adults)	
Age at death without treatment	Both patients will die at the same age	
Life expectancy without treatment (from today)	1 year	1 year
Health without treatment (in final year of life)	50%	50%
Gain from treatment	No life extension Restore to 100% health	+ 1 year No health improvement

Questions [same for all scenarios]

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

- ☐ I would prefer the health service to treat Patient A
- ☐ I have no preference
- ☐ I would prefer the health service to treat Patient B

Which of the following statements reflect the reason(s) for your answer to question 1?

(tick all boxes that apply)

- ☐ My choice delivers the largest benefit
- ☐ My choice is the most fair
- ☐ My choice delivers the benefit today rather than far away in the future
- ☐ My choice benefits the patient who is closest to death
- ☐ My choice benefits the patient who has longer left to live
- ☐ My choice benefits the patient with less time to prepare for death
- ☐ My choice benefits the patient who can make the most out of their remaining time
- ☐ My choice benefits the patient who is worse off
- ☐ My choice benefits the patient who is younger today
- ☐ My choice benefits the patient who is older today
- ☐ My choice benefits the patient who will die at a younger age
- ☐ My choice benefits the patient who will die at an older age
- ☐ I think that it is better to improve health than to extend life in this situation
- ☐ I think that it is better to extend life than to improve health in this situation
- ☐ I think that both patients are equally deserving of treatment
- ☐ I think that it is unfair to choose between the patients
- ☐ I am unwilling to choose between the patients
- ☐ None of the above

Appendix 7 Empirical study 2 information sheet

1. **Research Project Title:**

Health care priority setting preference project

2. **Invitation paragraph**

You are being invited to take part in a research project. Before you decide whether you wish to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Take time to decide whether or not you wish to take part. Thank you for reading this.

3. **What is the project's purpose?**

The purpose of the project is to find out what the general public thinks about a range of hypothetical scenarios where health care decision makers have to choose which types of treatment to allocate funding to. Better understanding of public preferences will help organisations such as the National Institute for Health and Clinical Excellence (NICE) to make appropriate resource allocation decisions.

4. **Why have I been chosen?**

We are seeking adult participants from the general population in England and Wales. We intend to recruit about 50 participants in total.

5. **Do I have to take part?**

It is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep (and be asked to sign a consent form) and you can still withdraw at any time without it affecting any benefits that you are entitled to in any way. If you decide to stop, then any information that you have provided will be destroyed. You do not have to give a reason for not taking part.

6. **What will happen to me if I take part?**

If you agree to take part, you will complete an interview survey that should last for no more than 30 minutes. You will be interviewed by a researcher, who will ask you a series of questions which require you to compare hypothetical scenarios in which a health care decision maker must allocate resources to one of two treatments for ill health. A paper questionnaire will be used to provide illustrations of the scenarios and to record your responses. You will also be asked some questions about your thoughts and opinions about the exercise, as well as some questions about your experience of ill health. You will only be asked to participate in one interview.

7. **What do I have to do?**

You will be asked to answer a series of questions involving hypothetical scenarios. There are no right or wrong answers – we are simply seeking your views.

8. **What are the possible disadvantages and risks of taking part?**

Some participants may feel uncomfortable when asked to think about scenarios involving illness and death. However, previous research in this area has shown that participants are generally interested and engaged when taking part in these types of exercises.

9. **What are the possible benefits of taking part?**

All participants will be given a small cash payment for agreeing to take part in the study. You will also be contributing to research that will help health care decision makers to better understand the preferences of members of the general public.

10. **What if something goes wrong?**

Should you wish to raise a complaint regarding your treatment by researchers or any other aspect of the study, you should contact Professor Aki Tsuchiya (a.tsuchiya@sheffield.ac.uk), who will follow up on your complaint immediately. However, should you feel that your complaint has not been handled to your satisfaction, you can contact the University's Registrar and Secretary (telephone: 0114 222 1100; email: registrar@sheffield.ac.uk).

11. **Will my taking part in this project be kept confidential?**

All information which is collected about you during the course of the research will be kept strictly confidential. All survey responses will be anonymised, so you will not be identified in any reports or publications.

12. **What will happen to the results of the research project?**

The results of the project will be written up in a report for the National Institute for Health and Clinical Excellence (NICE), and will be published in academic journals and presented at conferences. You will not be identified in any reports or publications. The anonymised data collected during the course of the project may be used for additional or subsequent research and analysis.

13. **Who is organising and funding the research?**

The project is being organised by Allan Wailoo, Aki Tsuchiya, and Koonal Shah, of the University of Sheffield's' School of Health and Related Research. It has been funded by the National Institute for Health and Clinical Excellence. The University has a contract with Accent, an experienced market research agency, to carry out the interviews for this project.

14. **Who has ethically reviewed the project?**

The project has been reviewed by the School of Health and Related Research Ethics Committee at the University of Sheffield.

15. **Contact for further information**

For information regarding participant recruitment and co-ordination of the interview programme, please contact Koonal Shah (k.k.shah@sheffield.ac.uk; 07920 496832). For any other issues relating to the research project, please contact Professor Aki Tsuchiya (a.tsuchiya@sheffield.ac.uk; 0114 222 0710).

If you wish to seek further information about the topics covered in this project, you may find it helpful to get in touch with the Dying Matters Coalition, a group set up by the National Council for Palliative Care. You can find information, resources and details of organisations providing support and counselling on their website, http://www.dyingmatters.org.

16. **Will I be recorded, and how will the recorded media be used?**

No, we will not take any audio or video recording of the interviews.

Appendix 8 Empirical study 3 instructions for standard tasks

--

Thank you for agreeing to take part in this research project.

The main survey consists of 12 questions about hypothetical scenarios. Once you have completed these questions, you will be asked some further questions about yourself.

--

We are going to show you some hypothetical scenarios involving patients who are affected by illness. We will use the survey to ask you which patients you think the health service should treat.

--

Illnesses and medical treatments affect people's health and how long they live.

Different illnesses affect people's health and how long they live in different ways; and different treatments offer different types of benefits.

We are going to use pictures to show these differences in illnesses and treatments. On the following pages, we will explain how the pictures work.

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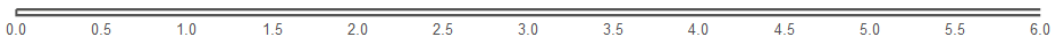

We can represent time with a line starting from 0 and going on to the right into the future.

Let's suppose that someone will live for 6 years from today. This can be shown by the line going from 0 years to 6 years.

--

We can also show how good someone's health is using a health scale, where **'dead' is 0%** and **'full health' is 100%**.

Of course, full health for a young person may be different from full health for an elderly person. But to keep things simple, we show full health for everyone as 100%.

Someone who has health problems would have a health level of less than 100%.

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**.

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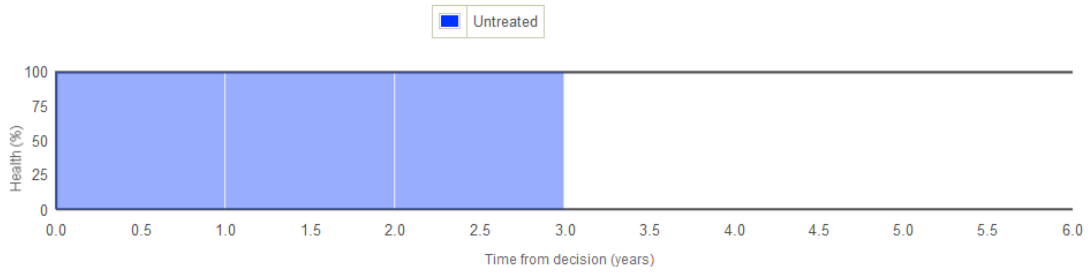

The blue area shows someone with an illness that gives the patient **3 years to live** from today, without treatment. This is shown by the end of the area at 3 years.

Note that the level of health is 100%, which represents full health. This means that although the illness leads to death in 3 years, it does not affect the patient's general health during those 3 years.

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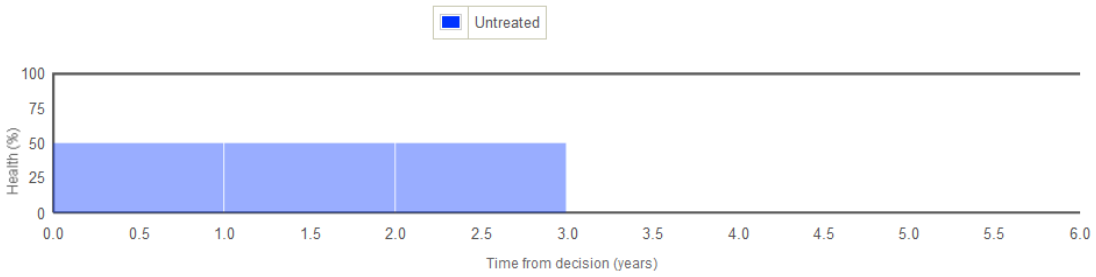

This blue area shows another illness. Without treatment, the patient shown here will live for 3 years in **50% health**, and then they will die.

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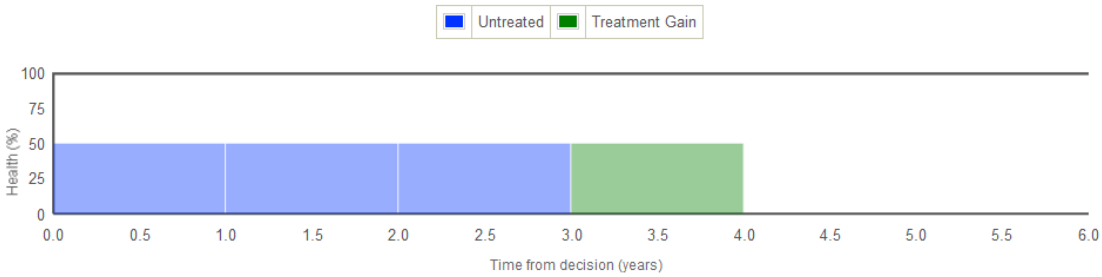

The green area shows a treatment for that illness. The treatment shown here gives the patient an **extra 1 year of life** at the same level of health (50%).

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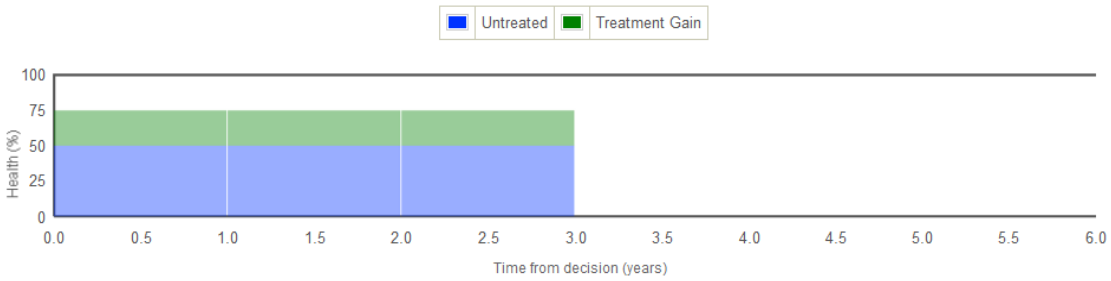

This treatment restores some of the patient's health (to 75%) but does not extend their life.

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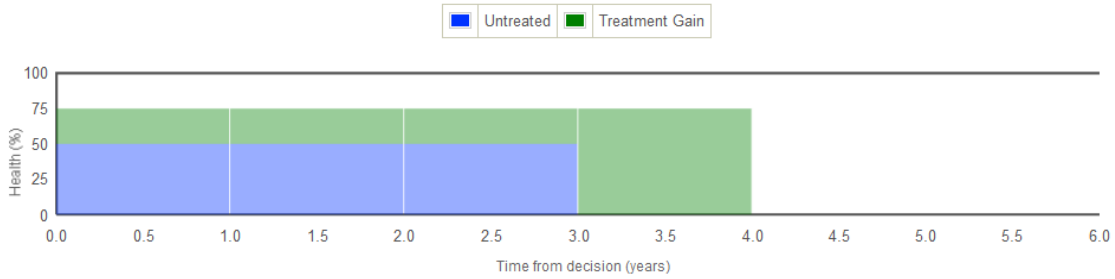


This treatment improves the patient's health to 75% AND gives them an extra 1 year of life.

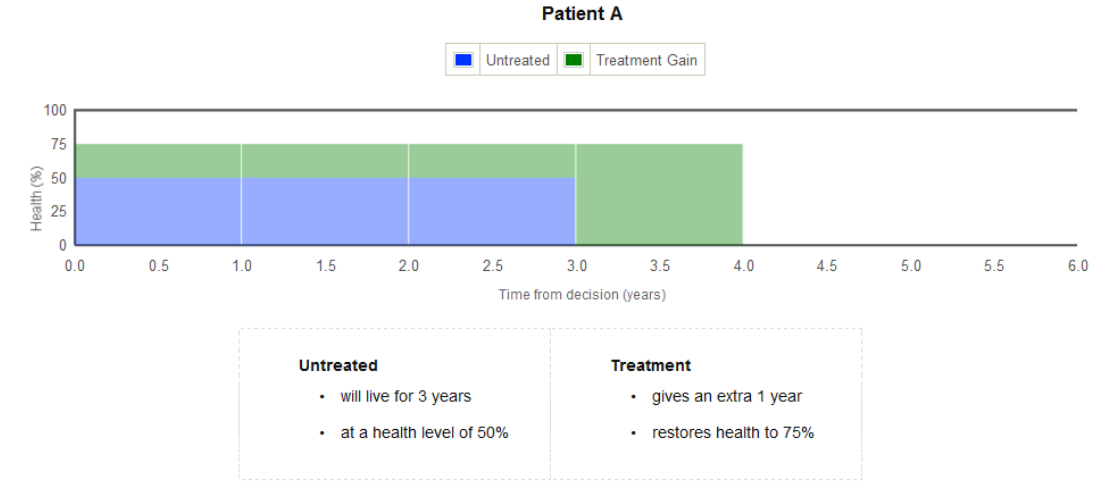
--

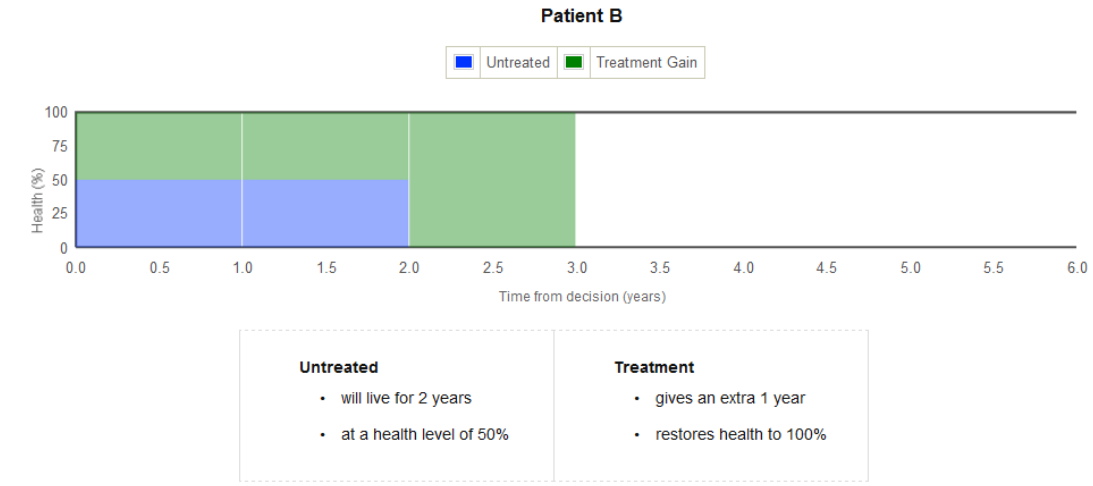
In the following questions you will asked to consider the situations of 2 hypothetical patients - patient A and patient B.

The patients will have different illnesses that affect their level of health and length of life in different ways.

The treatments available will also affect their health and length of life in different ways.

Scroll down to see an example of how the information about patient A and patient B will be shown in the questions.





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No other information about the patients is available, except that they are **both adults**. You should therefore consider them to be equal in all other respects.

We want you to assume that the health service has only enough funds to treat one of the two patients, and that **there are no alternative treatments available**.

Furthermore, the nature of the illnesses is such that further treatment will not be possible if either patient is not treated today - this is the only opportunity for treatment.

We want you to tell us which patient you think should be treated.

There are no right or wrong answers - we are simply seeking your view.

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Appendix 9 Empirical study 3 instructions for extension tasks

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The next 2 questions will require you to consider slightly different scenarios.

Just as before, the patients will have different illnesses that affect their health and length of life in different ways.

But in these scenarios, **one of the patients has known about their illness for some time** while the other patient has only just learned of their illness.

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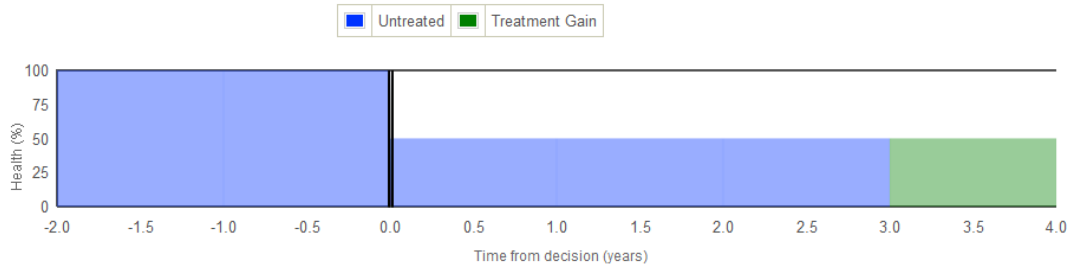

This patient was told **2 years ago** that they have **5 years to live**. This means that from today, they have **3 years to live**, unless they receive treatment.

Note that the blue area to the left of 0 years is at 100% health. This means that up until today, the illness has not affected the patient's general health.

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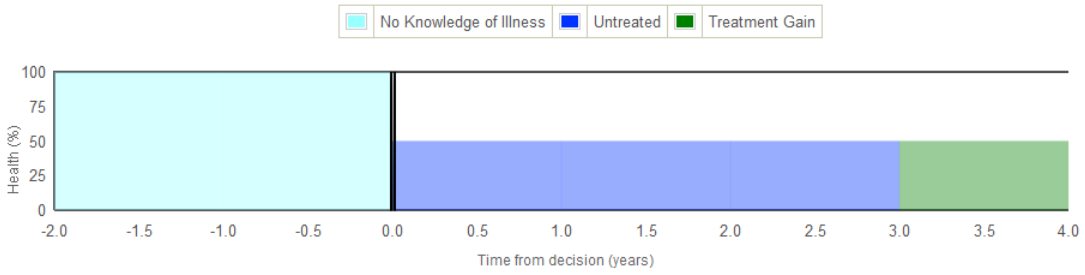


This patient has **just been told about their illness**. From today, they will live for 3 years before dying, unless they receive treatment.

The light blue area to the left of 0 years shows that the patient had **no knowledge of their illness** up until today.

--

Once again, we want you to tell us which patient you think should be treated.

--

Appendix 10 Empirical study 3 information sheet

Health care priority setting preference project

You are being invited to take part in a research project. Before you decide whether you wish to take part, it is important for you to understand why the research is being done and what it will involve. Please read the following information carefully and discuss it with others if you wish. Take time to decide whether or not you wish to take part. Thank you for reading this.

What is the project's purpose?

The purpose of the project is to find out what the general public thinks about a range of hypothetical scenarios where the health service has to choose which types of treatment to allocate funding to. Better understanding of public preferences will help organisations such as the NHS to make decisions about which treatments to provide.

Why have I been chosen?

We are seeking to survey around 4,000 members of the general public.

Do I have to take part?

It is up to you to decide whether or not to take part. If you do decide to take part you will be asked to complete an informed consent form and you can still withdraw at any time. If you decide to stop, then any information that you have provided will be destroyed. You do not have to give a reason for not taking part.

What will happen to me if I take part?

If you agree to take part, you will complete an online survey. The survey will involve looking at hypothetical scenarios in which a health care decision maker must allocate resources to one of two treatments for ill health. You will also be asked some questions about yourself.

What do I have to do?

You will be asked to answer a series of questions involving hypothetical scenarios. There are no right or wrong answers – we are simply seeking your views.

What are the possible disadvantages and risks of taking part?

Some participants may feel uncomfortable when asked to think about scenarios involving illness and death. However, previous research in this area has shown that participants are generally interested and engaged when taking part in these types of exercises. Remember – you are free to withdraw from participating at any time.

What are the possible benefits of taking part?

You will be contributing to research that will help health care decision makers to better understand the preferences of members of the general public.

What if something goes wrong?

Should you wish to raise a complaint about any aspect of the study, please send this to isabel@valuedopinions.co.uk.

Will my taking part in this project be kept confidential?

All information which is collected about you during the course of the research will be kept strictly confidential. All survey responses will be anonymised, so you will not be identified in any reports or publications.

What will happen to the results of the research project?

The results of the project will be written up in a report for the National Institute for Health and Clinical Excellence (NICE), and will be published in academic journals and presented at conferences. You will not be identified in any reports or publications. The anonymised data collected during the course of the project may be used for additional or subsequent research and analysis.

Who is organising and funding the research?

The project is being organised by Allan Wailoo, Aki Tsuchiya and Koonal Shah, of the University of Sheffield's School of Health and Related Research. It has been funded by NICE.

Who has ethically reviewed the project?

The project has been reviewed by the School of Health and Related Research Ethics Committee at the University of Sheffield.

Contact for further information

For further information about this survey, please contact Professor Aki Tsuchiya (a.tsuchiya@sheffield.ac.uk; 0114 222 0710).

If you wish to seek further information about the topics covered in this project, you may find it helpful to get in touch with the Dying Matters Coalition, a group set up by the National Council for Palliative Care. You can find information, resources and details of organisations providing support and counselling on their website, http://www.dyingmatters.org. To speak to someone for cancer support over the telephone, you may call the Macmillan Support Line: 0808 808 0000 (free).

Appendix 11 Empirical studies 1, 2, 3 ethics approval

Note: The letter below refers to the approval of empirical studies 1 and 2. Empirical study 3 was approved via email by the Chair of the ScHARR Ethics Committee (Dr Jennifer Burr) on the basis that it was the next phase of the project that had already been approved by the ScHARR Ethics Committee (Burr, J., 2011, personal communication, 12 December).

The University Of Sheffield.

Cheryl Oliver
Ethics Committee Administrator

Regent Court
30 Regent Street
Sheffield S1 4DA
Telephone: +44 (0) 114 2220871
Fax: +44 (0) 114 272 4095 (non confidential)
Email: c.a.oliver@sheffield.ac.uk

Our ref: 0461/CAO

25 July 2011

Koonal Shah
ScHARR

Dear Koonal

Valuing health at the end of life: simple choice study (lay title: Health care priority setting preference study)

Thank you for submitting the above research project for approval by the ScHARR Research Ethics Committee. On behalf of the University Chair of Ethics who reviewed your project, I am pleased to inform you that on 25 July 2011 the project was approved on ethics grounds, on the basis that you will adhere to the documents that you submitted for ethics review.

The research must be conducted within the requirements of the hosting/employing organisation or the organisation where the research is being undertaken.

If during the course of the project you need to deviate significantly from the documents you submitted for review, please inform me since written approval will be required. Please also inform me should you decide to terminate the project prematurely.

Yours sincerely

Cheryl Oliver
Ethics Committee Administrator

Appendix 12 Empirical study 3 estimated utility score and predicted probability of choice for all profiles

Table 0-6. Estimated utility score and predicted probability of choice for all 110 profiles (complete version of Table 5-6)

Rank	LE without treatment (mths)	QOL without treatment (%)	LE gain (mths)	QOL gain (%)	QALYs without treatment	QALYs gained	Utility	Prob.	Cumul. Prob.
1	60	50	12	50	2.500	3.500	4.3445	0.1351	0.1351
2	36	50	12	50	1.500	2.500	3.6380	0.0667	0.2018
3	12	50	12	50	0.500	1.500	3.3041	0.0477	0.2495
4	24	50	12	50	1.000	2.000	3.2848	0.0468	0.2964
5	60	50	6	50	2.500	3.000	3.0554	0.0372	0.3336
6	3	50	12	50	0.125	1.125	3.0392	0.0366	0.3702
7	3	100	12	0	0.250	1.000	2.7498	0.0274	0.3976
8	12	100	12	0	1.000	1.000	2.6977	0.0260	0.4237
9	60	50	12	25	2.500	2.000	2.5973	0.0235	0.4472
10	60	50	3	50	2.500	2.750	2.4109	0.0195	0.4668
11	12	50	12	25	0.500	1.000	2.4022	0.0194	0.4861
12	36	50	6	50	1.500	2.000	2.3490	0.0184	0.5045
13	36	50	12	25	1.500	1.500	2.3135	0.0177	0.5222
14	3	50	12	25	0.125	0.813	2.2958	0.0174	0.5396
15	24	100	12	0	2.000	1.000	2.2557	0.0167	0.5564
16	60	50	2	50	2.500	2.667	2.1961	0.0158	0.5721
17	36	100	12	0	3.000	1.000	2.1862	0.0156	0.5878
18	24	50	12	25	1.000	1.250	2.1716	0.0154	0.6031
19	60	100	12	0	5.000	1.000	2.0474	0.0136	0.6167
20	12	50	6	50	0.500	1.000	2.0150	0.0132	0.6299
21	24	50	6	50	1.000	1.500	1.9957	0.0129	0.6428
22	60	50	1	50	2.500	2.583	1.9812	0.0127	0.6555
23	60	50	0	50	2.500	2.500	1.7664	0.0103	0.6658
24	3	50	6	50	0.125	0.625	1.7501	0.0101	0.6759
25	36	50	3	50	1.500	1.750	1.7045	0.0096	0.6855
26	60	50	6	25	2.500	1.625	1.6535	0.0092	0.6947
27	3	100	6	0	0.250	0.500	1.5527	0.0083	0.7029
28	3	50	12	0	0.125	0.500	1.5525	0.0083	0.7112
29	12	100	6	0	1.000	0.500	1.5006	0.0079	0.7191
30	12	50	12	0	0.500	0.500	1.5004	0.0079	0.7270

Rank	LE without treatment (mths)	QOL without treatment (%)	LE gain (mths)	QOL gain (%)	QALYs without treatment	QALYs gained	Utility	Prob.	Cumul. Prob.
31	36	50	2	50	1.500	1.667	1.4896	0.0078	0.7347
32	12	50	6	25	0.500	0.625	1.4585	0.0075	0.7423
33	12	50	3	50	0.500	0.750	1.3705	0.0069	0.7492
34	36	50	6	25	1.500	1.125	1.3697	0.0069	0.7561
35	3	50	6	25	0.125	0.438	1.3520	0.0068	0.7629
36	24	50	3	50	1.000	1.250	1.3512	0.0068	0.7696
37	36	50	1	50	1.500	1.583	1.2748	0.0063	0.7759
38	24	50	6	25	1.000	0.875	1.2278	0.0060	0.7819
39	60	50	3	25	2.500	1.438	1.1816	0.0057	0.7876
40	24	50	2	50	1.000	1.167	1.1364	0.0055	0.7931
41	3	50	3	50	0.125	0.375	1.1056	0.0053	0.7984
42	36	50	0	50	1.500	1.500	1.0599	0.0051	0.8034
43	24	100	6	0	2.000	0.500	1.0587	0.0051	0.8085
44	24	50	12	0	1.000	0.500	1.0584	0.0051	0.8135
45	60	50	2	25	2.500	1.375	1.0243	0.0049	0.8184
46	36	100	6	0	3.000	0.500	0.9892	0.0047	0.8231
47	36	50	12	0	1.500	0.500	0.9890	0.0047	0.8278
48	12	50	3	25	0.500	0.438	0.9866	0.0047	0.8325
49	3	100	3	0	0.250	0.250	0.9542	0.0046	0.8371
50	3	50	6	0	0.125	0.250	0.9539	0.0046	0.8417
51	24	50	1	50	1.000	1.083	0.9215	0.0044	0.8461
52	12	100	3	0	1.000	0.250	0.9021	0.0043	0.8504
53	12	50	6	0	0.500	0.250	0.9019	0.0043	0.8547
54	36	50	3	25	1.500	0.938	0.8978	0.0043	0.8590
55	3	50	3	25	0.125	0.250	0.8801	0.0042	0.8632
56	60	50	1	25	2.500	1.313	0.8670	0.0042	0.8674
57	60	100	6	0	5.000	0.500	0.8503	0.0041	0.8715
58	60	50	12	0	2.500	0.500	0.8501	0.0041	0.8756
59	12	50	2	50	0.500	0.667	0.7832	0.0038	0.8795
60	24	50	3	25	1.000	0.688	0.7559	0.0037	0.8832
61	36	50	2	25	1.500	0.875	0.7405	0.0037	0.8869
62	60	50	0	25	2.500	1.250	0.7097	0.0036	0.8904
63	24	50	0	50	1.000	1.000	0.7067	0.0036	0.8940
64	3	50	3	0	0.125	0.125	0.6547	0.0034	0.8974

Rank	LE without treatment (mths)	QOL without treatment (%)	LE gain (mths)	QOL gain (%)	QALYs without treatment	QALYs gained	Utility	Prob.	Cumul. Prob.
65	12	50	3	0	0.500	0.125	0.6026	0.0032	0.9006
66	24	50	2	25	1.000	0.625	0.5986	0.0032	0.9038
67	36	50	1	25	1.500	0.813	0.5832	0.0031	0.9069
68	12	50	1	50	0.500	0.583	0.5683	0.0031	0.9100
69	3	50	2	50	0.125	0.292	0.5182	0.0029	0.9129
70	24	100	3	0	2.000	0.250	0.4601	0.0028	0.9157
71	24	50	6	0	1.000	0.250	0.4599	0.0028	0.9185
72	12	50	2	25	0.500	0.375	0.4567	0.0028	0.9213
73	24	50	1	25	1.000	0.563	0.4413	0.0027	0.9240
74	36	50	0	25	1.500	0.750	0.4259	0.0027	0.9267
75	36	100	3	0	3.000	0.250	0.3907	0.0026	0.9293
76	36	50	6	0	1.500	0.250	0.3904	0.0026	0.9319
77	3	100	2	0	0.250	0.167	0.3822	0.0026	0.9344
78	12	50	0	50	0.500	0.500	0.3535	0.0025	0.9369
79	3	50	2	25	0.125	0.188	0.3503	0.0025	0.9394
80	12	100	2	0	1.000	0.167	0.3301	0.0024	0.9419
81	3	50	1	50	0.125	0.208	0.3034	0.0024	0.9442
82	12	50	1	25	0.500	0.313	0.2994	0.0024	0.9466
83	24	50	0	25	1.000	0.500	0.2840	0.0023	0.9489
84	24	100	2	0	2.000	0.167	0.2606	0.0023	0.9512
85	60	100	3	0	5.000	0.250	0.2518	0.0023	0.9535
86	60	50	6	0	2.500	0.250	0.2515	0.0023	0.9557
87	3	50	1	25	0.125	0.125	0.1930	0.0021	0.9578
88	36	100	2	0	3.000	0.167	0.1912	0.0021	0.9600
89	3	100	1	0	0.250	0.083	0.1827	0.0021	0.9621
90	3	50	2	0	0.125	0.083	0.1824	0.0021	0.9642
91	24	50	3	0	1.000	0.125	0.1606	0.0021	0.9662
92	12	50	0	25	0.500	0.250	0.1421	0.0020	0.9683
93	12	100	1	0	1.000	0.083	0.1306	0.0020	0.9703
94	12	50	2	0	0.500	0.083	0.1303	0.0020	0.9723
95	36	50	3	0	1.500	0.125	0.0912	0.0019	0.9742
96	3	50	0	50	0.125	0.125	0.0886	0.0019	0.9761
97	3	50	1	0	0.125	0.042	0.0826	0.0019	0.9780
98	24	100	1	0	2.000	0.083	0.0611	0.0019	0.9799

Rank	LE without treatment (mths)	QOL without treatment (%)	LE gain (mths)	QOL gain (%)	QALYs without treatment	QALYs gained	Utility	Prob.	Cumul. Prob.
99	24	50	2	0	1.000	0.083	0.0609	0.0019	0.9817
100	60	100	2	0	5.000	0.167	0.0523	0.0018	0.9836
101	3	50	0	25	0.125	0.063	0.0357	0.0018	0.9854
102	12	50	1	0	0.500	0.042	0.0306	0.0018	0.9872
103	36	100	1	0	3.000	0.083	-0.0083	0.0017	0.9889
104	36	50	2	0	1.500	0.083	-0.0086	0.0017	0.9907
105	24	50	1	0	1.000	0.042	-0.0389	0.0017	0.9924
106	60	50	3	0	2.500	0.125	-0.0477	0.0017	0.9940
107	36	50	1	0	1.500	0.042	-0.1083	0.0016	0.9956
108	60	100	1	0	5.000	0.083	-0.1472	0.0015	0.9971
109	60	50	2	0	2.500	0.083	-0.1475	0.0015	0.9986
110	60	50	1	0	2.500	0.042	-0.2472	0.0014	1.0000

Appendix 13 Empirical study 3 analysis of respondent choice strategies and background characteristics

The overall results of empirical study 3 indicate that choices about which patient to treat are influenced more by the sizes of the health gains achievable from treatment than by patients' life expectancy or quality of life in absence of treatment. However, whilst this conclusion may reflect the 'average' view of the sample, it is likely that a variety of different strategies were used by different groups of respondents when completing the choice tasks. It may be that whilst many respondents support a QALY-maximisation type objective, there are other groups of respondents who consistently prefer to treat those who are worse off without treatment, or who advocate a mixture of the two approaches. It has also been observed that respondents in stated preference studies often fail to make trade-offs between attributes, instead basing their choices on simple rules or heuristics (Araña *et al.*, 2008). The additional analysis described in this appendix seeks to define subgroups of respondents according to the nature of their choices, and to shed light on some of the potential determinants of belonging to one or other of these subgroups.

Twelve different choice strategies that respondents might follow when faced with these sorts of choice sets were identified *a priori* (Table 0-7). The first six are concerned with choosing the profile associated with larger health gains from treatment; and the final six are concerned with choosing the profile associated with poorer health without treatment; all of these are examples of very simple decision rules that respondents might adopt. One would expect an advocate of NICE's policy of giving higher priority to life-extending end of life treatments to consistently follow strategy 8 (choose patient with less life expectancy without treatment) and strategy 2 (choose patient with larger life expectancy gain).

Choice strategies 4, 5 and 6 refer to choice sets in which both patients have a 'similar' number of QALYs without treatment. Likewise, choice strategies 10, 11 and 12 refer to choice sets in which both patients gain a 'similar' number of QALYs from treatment. The former are defined as instances where the difference between the better off patient and the worse off patient (in terms of their health without treatment) is less than or equal to 0.750 QALYs. The latter are defined as instances where the difference between the larger gain and the smaller gain is less than or equal to 0.917 QALYs. The reason why these particular cut-offs were used was a practical one – it meant that the same number of choice sets were captured by either rule (in 86 of the 160 choice sets, the patients had similar QALYs without treatment; likewise, in 86 of the 160 choice sets, the treatments offered similar QALY gains). The general results below are not sensitive to the choice of cut-offs used.

For each of the 160 choice sets, it was determined which choice (if any) would be consistent with each of the 12 strategies. The 10 choice sets faced by each individual respondent was then examined in order to determine whether or not their actual choices were consistent with each of the strategies. All 3,969 respondents faced the possibility of following or not following each of the 12 strategies on at each one occasion. The experimental design ensured that the patients always differed in terms of at least two of the four attributes. Hence, it was possible to follow or not follow several of the 12 strategies in any given choice set.

The third column in Table 0-7 shows the overall proportion of choices made by respondents that were consistent with each strategy when that strategy was

possible. A value of 1 in this column would indicate that on every occasion that respondents faced a choice set in which it was possible to follow the relevant strategy, they did indeed follow that strategy. The fourth, fifth and sixth columns show the number and proportion of respondents who never, sometimes and always followed each strategy, respectively. The proportions were calculated by using as the denominator the total number of respondents who faced at least one choice set in which it was possible to follow the relevant strategy. To 'never follow' a given strategy means to always follow the opposite strategy – for example, a respondent who always chose to treat the patient with greater QALYs without treatment can be said to have never followed strategy 7. Clearly, these statistics become less meaningful when the number of choice sets in which it was possible to follow a given strategy is very small (for example, 12.6% and 25.1% of respondents faced only one choice set in which it was possible to follow strategies 11 and 12, respectively).

Table 0-7: Summary of different choice strategies and how consistently they were followed by respondents

Choice strategy		Min / mean / max number of choice sets faced by respondents in which strategy was available	Prop. choices made according to this strategy (when available)	n (prop.) respondents who *never* followed this strategy	n (prop.) respondents who *sometimes* followed this strategy	n (prop.) respondents who *always* followed this strategy
1	Choose patient with larger QALY gain	8 / 9.6 / 10	0.75	1 (0.000)	3,530 (0.889)	438 (0.110)
2	Choose patient with larger LE gain	4 / 5.2 / 7	0.69	20 (0.005)	3,405 (0.858)	544 (0.137)
3	Choose patient with larger QOL gain	8 / 8.3 / 9	0.70	2 (0.001)	3,640 (0.917)	327 (0.082)
4	Choose patient with larger QALY gain when both have similar QALYs without treatment	3 / 5.0 / 7	0.78	29 (0.007)	2,449 (0.617)	1491 (0.376)
5	Choose patient with larger LE gain when both have similar QALYs without treatment	3 / 4.0 / 5	0.71	32 (0.008)	3,106 (0.783)	831 (0.209)
6	Choose patient with larger QOL gain when both have similar QALYs without treatment	4 / 4.6 / 6	0.70	21 (0.005)	3,107 (0.783)	841 (0.212)
7	Choose patient with fewer QALYs without treatment	6 / 7.2 / 8	0.47	182 (0.046)	3,701 (0.932)	86 (0.022)
8	Choose patient with less LE without treatment	5 / 5.6 / 6	0.45	355 (0.089)	3,434 (0.865)	180 (0.045)
9	Choose patient with less QOL without treatment	3 / 3.6 / 4	0.54	276 (0.070)	3,074 (0.775)	619 (0.156)
10	Choose patient with fewer QALYs	2 / 4.1 / 6	0.51	457 (0.115)	3,051 (0.769)	461 (0.116)

Choice strategy		Min / mean / max number of choice sets faced by respondents in which strategy was available	Prop. choices made according to this strategy (when available)	n (prop.) respondents who *never* followed this strategy	n (prop.) respondents who *sometimes* followed this strategy	n (prop.) respondents who *always* followed this strategy
	without treatment when both gain similar QALYs from treatment					
11	Choose patient with less LE without treatment when both gain similar QALYs from treatment	1 / 2.9 / 4	0.52	891 (0.224)	2,101 (0.529)	977 (0.246)
12	Choose patient with less QOL without treatment when both gain similar QALYs from treatment	1 / 2.7 / 4	0.50	732 (0.184)	2,366 (0.596)	871 (0.219)

This analysis reinforces the finding that concern for achieving larger health gains generally trumps concerns about treating the patient who is most severely ill or closest to their end of life. Respondents chose to treat the worse off patient (or the patient with shorter life expectancy) in less than 50% of all instances (strategies 7 and 8). The propensity to choose to treat the worse off patient increases only very slightly when analysis is restricted to choice sets in which both patients gain similar QALYs from treatment (strategies 10 and 11).

Moreover, 891 respondents (22.4%) *never* chose strategy 11 (that is, they never chose to treat the patient with shorter life expectancy) even when the gains from treating that patient were similar to those from treating the patient with longer life expectancy. Hence, there is a sizeable number of respondents who appear to actively reject the concept of giving priority to those at the end of life. This is consistent with the findings of empirical study 2.

Although the majority of respondents support a variety of different objectives when making choices about which patient to treat, there appear to be subgroups of respondents who hold opposing views from each other. It may be possible to predict whether or not a given respondent holds a particular view about priority-setting using information about their sociodemographic or health background.

Table 0-8 summarises the background characteristics of subgroups of respondents who consistently followed certain choice strategies that are of particular interest for this study. The summary statistics suggest that these subgroups are similar to each other and to the full sample with respect to most of the characteristics. A notable exception is that of age – the respondents who always support giving priority to the severely ill and/or to those at the end of life are somewhat younger than the respondents who reject such strategies. Respondents who support giving priority to the patient with fewer QALYs without treatment have lower levels of life satisfaction than do respondents who reject such strategies (two-sample t-test; $p<0.05$).

Table 0-8: Characteristics of respondents who always/never followed selected choice strategies

Characteristic		Always follow strategy 7 (always choose patient with least QALYs without treatment) (n=86)	Never follow strategy 7 (never choose patient with least QALYs without treatment) (n=182)	Always follow strategy 8 (always choose patient with least LE without treatment) (n=180)	Never follow strategy 8 (never choose patient with least LE without treatment) (n=355)	Overall sample (n=3,969)
Age	Mean years	40.7	53.8	41.7	55.5	48.9
Gender	% female	57%	47%	60%	42%	51%
Social grade	Mean; 1=A, 6=DE	3.35	3.49	3.49	3.48	3.31
Children	% Yes	47%	34%	39%	33%	38%
Education past school leaving age	% Yes	85%	67%	82%	68%	78%
Education to degree level	% Yes	45%	38%	43%	38%	46%
General health	Mean; 1=Very good; 5=Very poor	2.00	2.00	2.06	2.17	2.06
Health limitations	Mean; 1=Limited a lot; 3=Not limited	2.72	2.65	2.63	2.50	2.61
Mobility	Mean; 1=No problems; 5=Extreme problems	1.15	1.38	1.29	1.55	1.41
Self-care	Mean; 1=No problems; 5=Extreme problems	1.07	1.08	1.07	1.18	1.13
Usual activities	Mean; 1=No problems; 5=Extreme problems	1.28	1.36	1.35	1.54	1.42
Pain/ discomfort	Mean; 1=No problems; 5=Extreme problems	1.45	1.67	1.57	1.89	1.72
Anxiety/ depression	Mean; 1=No problems; 5=Extreme problems	1.70	1.49	1.76	1.61	1.59
Health satisfaction	Mean; 0=Completely dissatisfied; 10=Completely satisfied	6.85	6.95	6.82	6.59	6.93
Life satisfaction	Mean; 0=Completely dissatisfied; 10=Completely satisfied	6.91	7.38	6.79	7.08	7.09
Experience of terminal illness	% Yes	60%	66%	60%	70%	66%

In order to identify the driving factor(s) behind respondents' membership of the 'always follow choice strategy 7' and 'never follow choice strategy 7' subgroups, the

data were modelled using multinomial logit regressions. The model included 'category' as the dependent variable (1 = respondent sometimes follows choice strategy 7; 2 = respondent always follows choice strategy 7; 3 = respondent never follows choice strategy 7) and all of the background characteristics as the individual-specific explanatory variables. Marginal effects were then computed to show the change in the probability of belonging to either category 2 (as opposed to categories 1 and 3) or category 3 (as opposed to categories 1 and 2) following a marginal change in one of the explanatory variables. The results are shown in Table 0-9 and Table 0-10.

Table 0-9: Marginal effects for category 2 (change in probability of belonging to the 'always follow choice strategy 7' subgroup (n=3,969)

Variable	Marginal effect	p-value
Age	-0.0002821	0.033
Gender	0.0016252	0.685
Social grade	0.0003300	0.826
Children	0.0010360	0.683
Education past school leaving age	-0.0068714	0.229
Education to degree level	0.0027102	0.518
General health	0.0005779	0.869
Health limitations	-0.0015464	0.769
Mobility	-0.0119115	0.027
Self-care	0.0051298	0.501
Usual activities	0.0003138	0.949
Pain / discomfort	-0.0061657	0.085
Anxiety / depression	0.0011895	0.639
Health satisfaction	-0.0024644	0.065
Life satisfaction	0.0005622	0.642
Experience of terminal illness	0.0004673	0.898

Table 0-10: Marginal effects for category 3 (change in probability of belonging to the 'never follow choice strategy 7' subgroup (n=3,969)

Variable	Marginal effect	p-value
Age	0.0009494	0.000
Gender	-0.0026001	0.674
Social grade	0.0031572	0.152
Children	0.0015610	0.734
Education past school leaving age	0.0128890	0.080
Education to degree level	0.0055896	0.431
General health	-0.0107651	0.055
Health limitations	0.0046483	0.581
Mobility	0.0015486	0.823
Self-care	-0.0093990	0.378
Usual activities	-0.0019084	0.806
Pain / discomfort	-0.0050063	0.357
Anxiety / depression	0.0003044	0.947
Health satisfaction	-0.0051820	0.013
Life satisfaction	0.0024120	0.237
Experience of terminal illness	0.0027552	0.648

For example, with respect to category 2, the marginal effect of age is -0.00028, and is statistically significant at the 5% level. This can be interpreted as follows: as age increases by one unit (year), the probability of *always* choosing the patient with fewest QALYs decreases by 0.00028. Similarly, with respect to category 3, the marginal effect of age is 0.00095, and is also statistically significant. This means that as age increases by one year, the probability of *never* choosing the patient with fewest QALYs increases by 0.00095. It is worth noting that although the marginal effect of age is statistically significant, it is small in practical terms. All else equal, even a 30-year increase in age would not be sufficient for a 1% change in the probability of always choosing the patient with fewest QALYs. Including an age squared explanatory variable in the model reduces the statistical significance of the marginal effects of age but does not affect their signs.

Other than age, it is worth noting that the marginal effect of the health satisfaction variable is negative and statistically significant at the 10% level with respect to both category 2 and category 3. The means that as health satisfaction increases by one unit, the probability of always choosing the patient with fewest QALYs decreases, as does the probability of never choosing the patient with fewest QALYs. Again, however, the actual changes in probability are very small.

The marginal effects of most of the other variables are not statistically significant. This suggests that the other background characteristics do not appear particularly likely to determine whether the respondent always or never follows the strategy of choosing the patient with fewest QALYs.

Table 0-11 and Table 0-12 use the same method to identify the driving factor(s) behind respondents' membership of the 'always follow choice strategy 8' and 'never follow choice strategy 8' subgroups.

Table 0-11: Marginal effects for category 2 (change in probability of belonging to the 'always follow choice strategy 8' subgroup (n=3,969)

Variable	Marginal effect	p-value
Age	-0.0006512	0.001
Gender	0.0107338	0.091
Social grade	0.0028863	0.213
Children	-0.0022585	0.598
Education past school leaving age	-0.0105657	0.220
Education to degree level	0.0054587	0.417
General health	0.0030304	0.591
Health limitations	-0.0107831	0.171
Mobility	-0.0021591	0.765
Self-care	-0.0165401	0.164
Usual activities	-0.0031122	0.676
Pain / discomfort	-0.0096205	0.081
Anxiety / depression	0.0053571	0.174
Health satisfaction	-0.0016267	0.471
Life satisfaction	-0.0001371	0.943
Experience of terminal illness	0.0030492	0.599

Table 0-12: Marginal effects for category 3 (change in probability of belonging to the 'never follow choice strategy 8' subgroup (n=3,969)

Variable	Marginal effect	p-value
Age	0.0022183	0.000
Gender	-0.0199488	0.018
Social grade	0.0039996	0.181
Children	0.0076284	0.220
Education past school leaving age	0.0152432	0.129
Education to degree level	0.0157259	0.100
General health	-0.0143061	0.061
Health limitations	-0.0011434	0.916
Mobility	-0.0046376	0.592
Self-care	0.0040344	0.714
Usual activities	-0.0004983	0.958

Variable	Marginal effect	p-value
Pain / discomfort	0.0057664	0.402
Anxiety / depression	0.0047326	0.412
Health satisfaction	-0.0056320	0.058
Life satisfaction	0.0012172	0.652
Experience of terminal illness	-0.0012765	0.880

Again, with respect to category 2, the marginal effect of age is negative and statistically significant; and with respect to category 3, the marginal effect of age is positive and statistically significant. This means that as age increases by one year, the probability of always choosing the patient with less life expectancy decreases while the probability of never choosing the patient with less life expectancy increases. Again, including an age squared explanatory variable in the model reduces the statistical significance of the marginal effects of age but does not affect their signs.

The marginal effects of most of the other variables are not statistically significant. An exception to this is the marginal effect of gender, which is positive with respect to category 2 and negative with respect to category 3. This means that as gender changes from male to female, the probability of always choosing the patient with less life expectancy increases whilst the probability of never choosing the patient with less life expectancy decreases.

The analysis in this appendix shows that the sample comprises multiple subgroups with clearly opposing views about priority-setting – for example, 8.9% of respondents always chose to treat the patient with longer life expectancy while 4.5% respondents always chose to treat the patient with shorter life expectancy. However, membership of these subgroups is not particularly well predicted by the observable characteristics on which information was collected in this study. This is consistent with the findings of the subgroup analysis that showed that excluding from the sample respondents with experience of close friends or family members with terminal illness, or respondents with responsibility with children, did not have a major impact on the regression results. There may be characteristics that are more difficult to observe (such as personal and cultural values) which are driving respondents' preferences and choice strategies.

Appendix 14 Empirical study 4 instructions for visual aid arm

Health priorities survey

Thank you for agreeing to take part in this research project.

The main survey consists of a series of questions about hypothetical scenarios. Once you have completed these questions, you will be asked some further questions about yourself and about your views about priorities for the health service.

→ Continue

We are going to show you some hypothetical scenarios involving patients who are affected by illness. We will use the survey to ask you which patients you think the health service should treat.

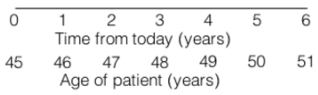
→ Continue

Illnesses and medical treatments affect people's health and how long they live.

Different illnesses affect people's health and how long they live in different ways; and different treatments offer different types of benefits.

We are going to use pictures to show these differences in illnesses and treatments. On the following pages, we will explain how the pictures work.

→ Continue



We can represent time with a line starting from 0 and going on to the right into the future.

Let's suppose that a 45 year old person will live for 6 years from today. This can be shown by the upper line going from 0 years to 6 years. The lower line shows the age of the patient.

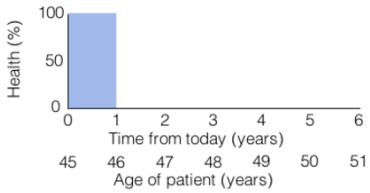
→ Continue

We can also show how good someone's health is using a 'health thermometer', where **dead is 0%** and **'full health' is 100%**.

Of course, full health for a young person may be different from full health for an older person. But to keep things simple, we show full health for everyone as 100%.
Someone who has health problems would have a health level of less than 100%.

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**.

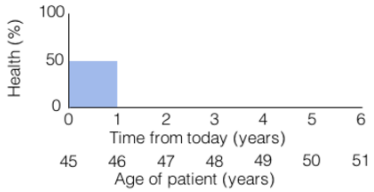
→ Continue



The blue box shows someone with an illness that gives the patient **1 year to live** from today, without treatment. This is shown by the end of the box at 1 year.

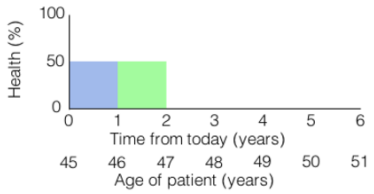
Note that the level of health is 100%, which represents full health. This means that although the illness leads to death in 1 year, it does not affect the patient's general health during that 1 year.

→ Continue



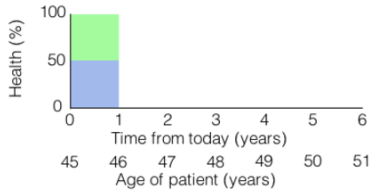
This blue box shows another illness. Without treatment, the patient shown here will live for 1 year **in 50% health**, and then they will die.

→ Continue



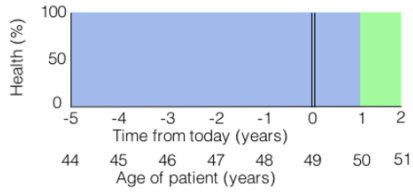
The green box shows a treatment for that illness. The treatment shown here gives the patient **an extra 12 months** of life at the same level of health (50%).

→ Continue



This treatment **restores the patient to full health** but does not extend their life.

→ Continue

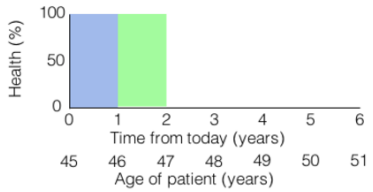


In some cases, the patient may have known about their illness for some time.

This patient was told **5 years ago** that they have **6 years to live**. This means that from today, they have **1 year to live**, unless they receive treatment.

Note that the blue box to the left of 0 years is at 100% health. This means that up until today, the illness has not affected the patient's general health.

→ Continue



This patient has **just been told about their illness**. From today, they will live for 1 year before dying, unless they receive treatment.

Note that there is no blue box to the left of 0 years. This shows that the patient had no knowledge of their illness up until today.

→ Continue

In the following questions you will be asked to consider the situations of two hypothetical patients – **patient A** and **patient B**.

The patients will have different illnesses that affect their health and length of life in different ways.

In some cases, the patients will have known about their illness for some time. In other cases, they will have just learned of their illness.

The treatments available will also affect their health and length of life in different ways.

You will be given information about the patients' ages. No other information about the patients is available. Please assume that the patients are similar to each other in all other respects.

→ Continue

→ Continue

There are no right or wrong answers – we are simply seeking your view.

We want you to tell us which patient you think should be treated.

We want you to imagine that the patient, if we have exactly as described – for example, there is no possibility that new treatments will be discovered.

The nature of the illnesses is such that further treatment will not be possible if either patient is not treated today – **this is the only opportunity for treatment**.

We want you to assume that the health service has only enough funds to treat one of the two patients, and that **there are no alternative treatments available**.

Appendix 15 Empirical study 4 instructions for no visual aid arm

Health priorities survey

Thank you for agreeing to take part in this research project.

The main survey consists of a series of questions about hypothetical scenarios. Once you have completed these questions, you will be asked some further questions about yourself and about your views about priorities for the health service.

→ Continue

→ Continue

We are going to show you some hypothetical scenarios involving patients who are affected by illness. We will use the survey to ask you which patients you think the health service should treat.

Illnesses and medical treatments affect people's health and how long they live.

Different illnesses affect people's health and how long they live in different ways; and different treatments offer different types of benefits.

→ Continue

We will describe how long a patient will live for by telling you their **life expectancy**, in years.

We will describe how good a patient's health is using percentages, where **dead is 0%** and **'full health' is 100%.**

Of course, full health for a young person may be different from full health for an older person. But to keep things simple, we describe full health for everyone as 100%.

Someone who has health problems would have a health level of less than 100%.

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**.

→ Continue

A patient may have an illness which affects their life expectancy but does not affect their general health.

Another patient may have an illness which affects both their life expectancy and their general health.

Similarly, the treatment for an illness may give patients extra months of life, or improve their general health, or both.

→ Continue

In some cases, the patient may have **known about their illness for some time**.

In other cases, the patient will have **just been told about their illness**, and will have had no knowledge of their illness up until today.

→ Continue

In the following questions you will be asked to consider the situations of two hypothetical patients – **patient A** and **patient B**.

The patients will have different illnesses that affect their health and length of life in different ways.

In some cases, the patients will have known about their illness for some time. In other cases, they will have just learned of their illness.

The treatments available will also affect their health and length of life in different ways.

You will be given information about the patients' ages. No other information about the patients is available. Please assume that the patients are similar to each other in all other respects.

→ Continue

We want you to assume that the health service has only enough funds to treat one of the two patients, and that **there are no alternative treatments available**.

The nature of the illnesses is such that further treatment will not be possible if either patient is not treated today – **this is the only opportunity for treatment**.

We want you to imagine that the patients' lives will be lived exactly as described – for example, there is no possibility that new treatments will be discovered.

We want you to tell us which patient you think should be treated.

There are no right or wrong answers – we are simply seeking your view.

→ Continue

Appendix 16 Empirical study 4 survey instrument

Note: These screenshots are taken from the forced choice, visual aid arm.

Scenario S1

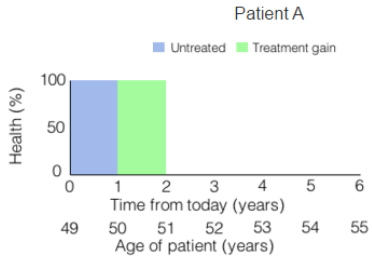
Consider two patients, patient A and patient B. Suppose that both patient A and patient B have just been diagnosed with illnesses.

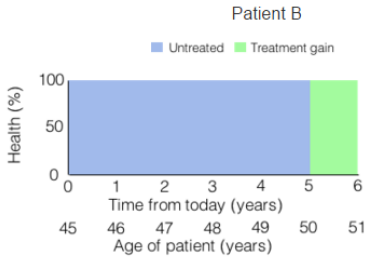
Patient A will live for 1 year, from today, before dying. Patient B will live for 5 years, from today, before dying. Both patients will die at the age of 50 as things stand.

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

✔ Submit answer

Scenario S2

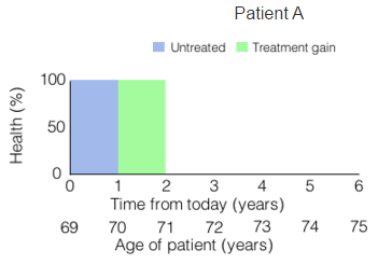
Consider two patients, patient A and patient B. Suppose that both patient A and patient B have just been diagnosed with illnesses.

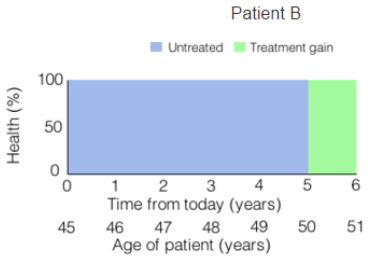
Patient A will live for 1 year, from today, before dying. Patient B will live for 5 years, from today, before dying. As things stand, patient A will die at the age of 70 and patient B will die at the age of 50.

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	69 years	45 years
Age at death without treatment	70 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

✔ Submit answer

Scenario S3

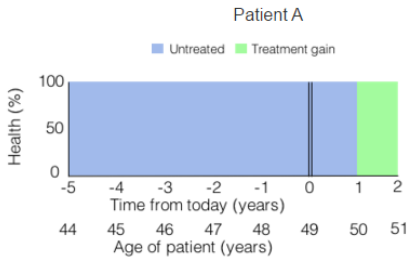
Consider two patients, patient A and patient B. Patient A has an illness which they were diagnosed with 5 years ago. Patient B has just been diagnosed with an illness.

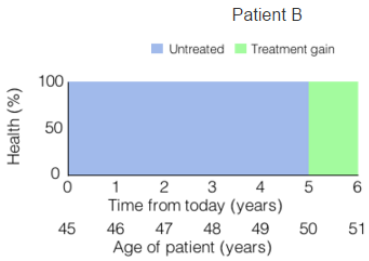
Patient A will live for 1 year, from today, before dying. Patient B will live for 5 years, from today, before dying. Both patients will die at the age of 50 as things stand.

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	Diagnosed 5 years ago (when aged 44 years)	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

✔ Submit answer

Scenario S4

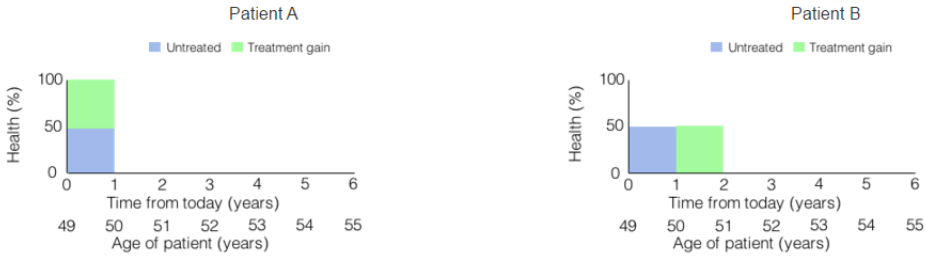
Consider two patients, patient A and patient B. Suppose that both patient A and patient B have just been diagnosed with illnesses.

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

As a result of their illnesses, both patients will experience a poorer level of general health – if left untreated, they will be in 50% health.

There is a treatment, which, if taken today, would restore patient A to full health. It would not affect patient A's life expectancy. Another treatment would, if taken today, extend the life of patient B by 12 months. It would not affect patient B's general level of health, so patient B's remaining life would be lived at 50% health.

	Patient A	**Patient B**
Age today	49 years	49 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	1 year
Health without treatment	50%	50%
Gain from treatment	No life extension + 50% health (restore to full health)	+ 12 months No health improvement



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

✔ Submit answer

Scenario S5

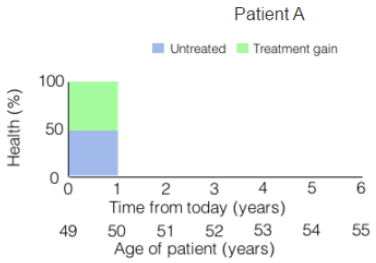
Consider two patients, patient A and patient B. Suppose that both patient A and patient B have just been diagnosed with illnesses.

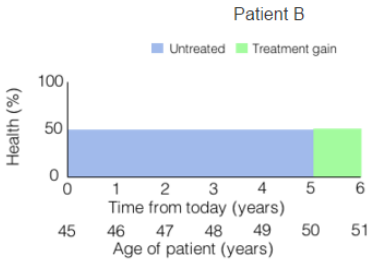
Patient A will live for 1 year, from today, before dying. Patient B will live for 5 years, from today, before dying. Both patients will die at the age of 50 as things stand.

As a result of their illnesses, both patients will experience a poorer level of general health – if left untreated, they will be in 50% health.

There is a treatment, which, if taken today, would restore patient A to full health. It would not affect patient A's life expectancy. Another treatment would, if taken today, extend the life of patient B by 12 months. It would not affect patient B's general level of health, so patient B's remaining life would be lived at 50% 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	50%	50%
Gain from treatment	No life extension + 50% health (restore to full health)	+ 12 months No health improvement





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

✔ Submit answer

Scenario S6

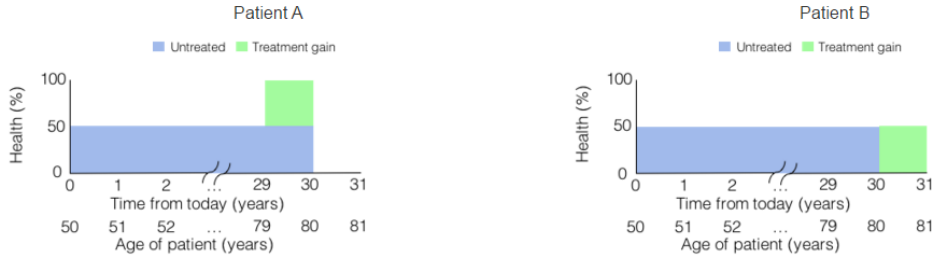
Consider two patients, patient A and patient B. Suppose that both patient A and patient B have just been diagnosed with illnesses.

As a result of their illnesses, both patients will experience a poorer level of general health – if left untreated, they will be in 50% health.

The illnesses do not affect how long the patients will live for. Both patients are the same age and will have a normal life expectancy of 80 years as things stand.

There is a treatment, which, if taken today, would restore patient A to full health in their final year of life (the preceding years would continue to be lived in 50% health). It would not affect patient A's life expectancy. Another treatment would, if taken today, extend the life of patient B by 12 months. It would not affect patient B's general level of health, so patient B's remaining life would be lived at 50% health.

	Patient A	**Patient B**
Age today	50 years	50 years
Age at death without treatment	80 years	80 years
Timing of diagnosis	Just been diagnosed	Just been diagnosed
Life expectancy without treatment (from today)	30 years	30 years
Health without treatment	50%	50%
Gain from treatment	No life extension + 50% health (restore to full health – final year of life only)	+ 12 months No health improvement



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

✔ Submit answer

Scenario S7

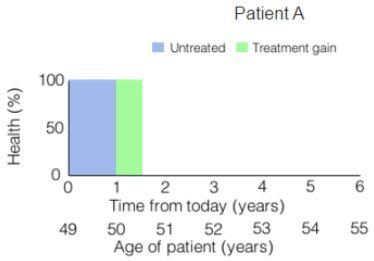
Consider two patients, patient A and patient B. Suppose that both patient A and patient B have just been diagnosed with illnesses.

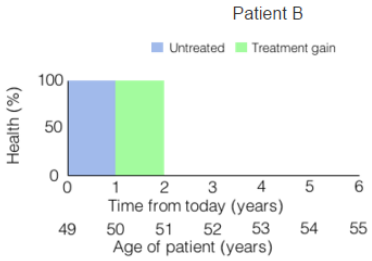
Patient A will live for 1 year, from today, before dying. Patient B will also live for 1 year, from today, before dying. Both patients will die at the age of 50 as things stand.

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

There is a treatment, which, if taken today, would extend the life of patient A by 6 months. Another treatment would, if taken today, extend the life of patient B by 12 months. Treatment would not affect either patient's general level of health.

	Patient A	**Patient B**
Age today	49 years	49 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	1 year
Health without treatment	100%	100%
Gain from treatment	+ 6 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

Submit answer

Follow-up question after having chosen a life-extending gain in a given scenario

Your choice was to treat patient B, 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**?

	Patient A	**Patient B**
Age today	69 years	45 years
Age at death without treatment	70 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	+ [ ] months

✔ Submit answer

Less than 1
1
2
3
4
5
6
7
8
9
10
11
12

Follow-up question after having chosen a QOL-improving gain in a given scenario

Your choice was to treat patient A, who would gain 50% health from treatment.

How much **smaller** would that 50% gain need to be for you to think that treating either patient would be **equally good**?

	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	50%	50%
Gain from treatment	No life extension + [ ] % health	+ 12 months No health improvement

✔ Submit answer

Less than 10
10
20
30
40
50

Follow-up question after having chosen smaller gain in scenario 7

Your choice was to treat patient A, who would gain 6 months from treatment.

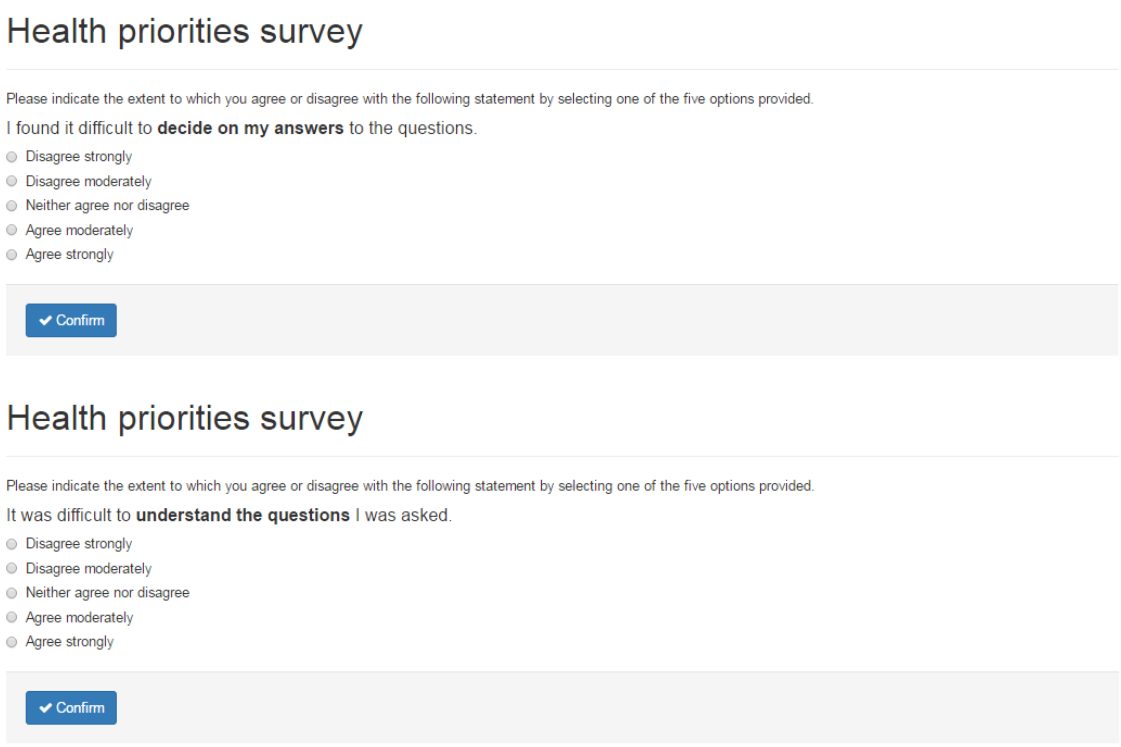
How much **shorter** would that 6 month gain need to be for you to think that treating either patient would be **equally good**?

	Patient A	**Patient B**
Age today	49 years	49 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	1 year
Health without treatment	100%	100%
Gain from treatment	+ [ ] months	+ 12 months

✔ Submit answer

Less than 1
1
2
3
4
5
6

Feedback / self-reported difficulty questions



Scenario 8 (individual perspective)

Health priorities survey

We are now going to ask you a question of a different sort. In the previous questions you were asked which of two patients should be treated, from the point of view of someone making decisions on behalf of the health service. However, we also want to know how you would respond when thinking about a situation that could affect you personally.

Now we want you to imagine that **you** could be one of the patients in need of treatment. Below two scenarios are presented, each of which describes a situation that could happen to you. Please suppose that there is a 50% chance that scenario A will occur and a 50% chance that scenario B will occur. Do not worry about whether the scenarios are realistic or not – you can help us to measure your preferences correctly by imagining that they would occur exactly as described.

Scenario A (50% chance of occurring)

You are 49 years old, and have just been diagnosed with illness A. As a result of illness A, your life expectancy is shortened – you will live for 1 year from the time of your diagnosis, and then you will die (aged 50 years). Illness A does not affect your general level of health – you will live in good health until you die.

Scenario B (50% chance of occurring)

You are 45 years old, and have just been diagnosed with illness B. As a result of illness B, your life expectancy is shortened – you will live for 5 years from the time of your diagnosis, and then you will die (aged 50 years). Illness B does not affect your general level of health – you will live in good health until you die.

We want you to choose between two treatments – one that is used to treat illness A (treatment A) and another that is used to treat illness B (treatment B).

Treatment A and treatment B both have identical effects. If taken at the time of diagnosis, these treatments will extend your life by 12 months. The treatments have no side effects. However, treatment A works only for illness A, and treatment B works only for illness B.

So, if scenario A were to occur, then treatment A would enable you to live for 2 years from the time of your diagnosis, after which you would die (aged 51 years).

And if scenario B were to occur, then treatment B would enable you to live for 6 years from the time of your diagnosis, after which you would die (aged 51 years).

	Scenario A	**Scenario B**
Age at diagnosis	49 years	45 years
Age at death without treatment	50 years	50 years
Life expectancy without treatment (from today)	1 year	5 years
Health without treatment	100%	100%
Gain from treatment (if treatment is available)	+ 12 months	+ 12 months

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?

- I would prefer treatment A to be available
- I have no preference (I do not mind which treatment is available)
- I would prefer treatment B to be available

✔ Confirm

Attitudinal questions

Health priorities survey

Please indicate the extent to which you agree or disagree with **the statement in bold** by selecting one of the five options provided.

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

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

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

- Disagree strongly
- Disagree moderately
- Neither agree nor disagree
- Agree moderately
- Agree strongly

✔ Confirm

Health priorities survey

Please indicate the extent to which you agree or disagree with **the statement in bold** by selecting one of the five options provided.

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

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

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

- Disagree strongly
- Disagree moderately
- Neither agree nor disagree
- Agree moderately
- Agree strongly

✔ Confirm

Health priorities survey

Please indicate the extent to which you agree or disagree with **the statement in bold** by selecting one of the five options provided.

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

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

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

- Disagree strongly
- Disagree moderately
- Neither agree nor disagree
- Agree moderately
- Agree strongly

✔ Confirm

Health priorities survey

Please indicate the extent to which you agree or disagree with **the statement in bold** by selecting one of the five options provided.

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.

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.

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.

- Disagree strongly
- Disagree moderately
- Neither agree nor disagree
- Agree moderately
- Agree strongly

✔ Confirm

Health priorities survey

Please indicate the extent to which you agree or disagree with **the statement in bold** by selecting one of the five options provided.

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.

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.

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.

- Disagree strongly
- Disagree moderately
- Neither agree nor disagree
- Agree moderately
- Agree strongly

✔ Confirm

Health priorities survey

Please indicate the extent to which you agree or disagree with **the statement in bold** by selecting one of the five options provided.

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.

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.

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.

- Disagree strongly
- Disagree moderately
- Neither agree nor disagree
- Agree moderately
- Agree strongly

✔ Confirm

Background questions

Health priorities survey

Are there any children (under 18 years) for whom you have responsibility?

- Yes
- No

✔ Confirm

Health priorities survey

Did your education continue after the minimum school leaving age?

- Yes
- No

✔ Confirm

Health priorities survey

Do you have a Degree or equivalent professional qualification?

- Yes
- No

✔ Confirm

Health priorities survey

How is your health in general?

- Very good
- Good
- Fair
- Poor
- Very poor

✔ Confirm

Health priorities survey

Are your day-to-day activities limited because of a health problem or disability which has lasted, or is expected to last, at least 12 months?

- Yes, limited a lot
- Yes, limited a little
- No

✔ Confirm

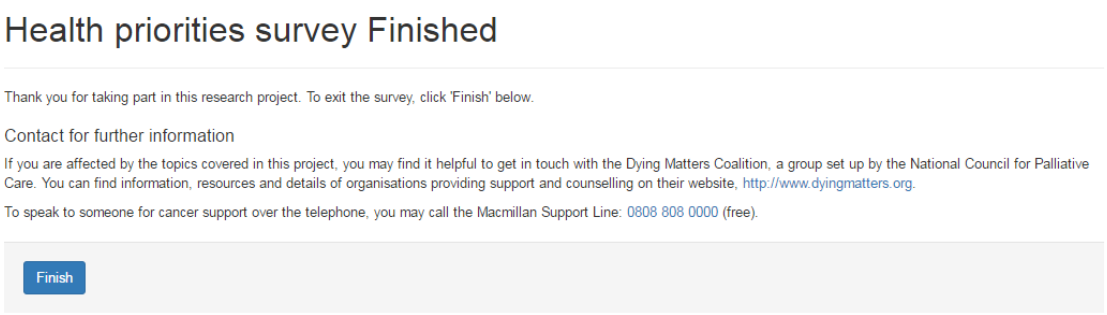
Health priorities survey

Do you have personal experience of a close friend or family member who has passed away following a period of illness (sometimes called terminal illness)?

- Yes
- No
- I would rather not answer this question

✔ Confirm

End screen


Health priorities survey Finished
Thank you for taking part in this research project. To exit the survey, click 'Finish' below.
Contact for further information
If you are affected by the topics covered in this project, you may find it helpful to get in touch with the Dying Matters Coalition, a group set up by the National Council for Palliative Care. You can find information, resources and details of organisations providing support and counselling on their website, http://www.dyingmatters.org.
To speak to someone for cancer support over the telephone, you may call the Macmillan Support Line: 0808 808 0000 (free).
Finish

Appendix 17 Empirical study 4 information sheet

Health priorities survey

Health priorities survey - Information Sheet

Health Priorities Survey / Principal Investigator: Professor Allan Wailoo Main study / Information Sheet / 21 December 2015

You are being invited to take part in a research project. This project is being carried out by researchers at the School of Health and Related Research, University of Sheffield. Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve. Please read the following information carefully and discuss it with others if you wish. Take time to decide whether or not you wish to take part.

1. What is the project's purpose?
The purpose of the project is to find out what the general public thinks about a range of hypothetical scenarios where health care decision makers have to choose which types of treatment to allocate funding to. Better understanding of public preferences will help organisations such as the National Institute for Health and Care Excellence (NICE) to decide how best to allocate limited health care resources.

2. Why have I been chosen?
We are seeking to survey around 2,400 members of the general public aged 18 years or older.

3. Do I have to take part?
It is up to you to decide whether or not to take part. If you decide to take part you will be asked to sign a consent form. You can withdraw at any time without any penalty to you. If you decide to stop, any information that you have provided will be destroyed. You do not have to give a reason for not taking part.

4. What will happen to me if I take part?
If you agree to take part, you will complete an online survey. The survey will involve looking at hypothetical scenarios in which a health care decision maker must allocate resources to one of two treatments for ill health. You will also be asked some questions about yourself and your views about priorities for the health service.

5. What do I have to do?
You will be asked to answer a series of questions involving hypothetical scenarios. There are no right or wrong answers – we are simply seeking your views.

6. What are the possible disadvantages and risks of taking part?
Some participants may feel uncomfortable when asked to think about scenarios involving illness and death. Remember – you are free to withdraw from participating at any time.

7. What are the possible benefits of taking part?
You will be contributing to research that will help health care decision makers to better understand the preferences of members of the general public.

8. What if something goes wrong?
Should you wish to raise a complaint regarding your treatment by researchers or any other aspect of the study, you should contact Professor Aki Tsuchiya (a.tsuchiya@sheffield.ac.uk; 0114 222 0710 / 0114 222 3422), who will follow up on your complaint immediately. However, should you feel that your complaint has not been handled to your satisfaction, you can contact Professor Jon Nicholl, Dean of the School of Health and Related Research (telephone: 0114 222 5453; email: j.nicholl@sheffield.ac.uk).

9. Will my taking part in this project be kept confidential?
All information which is collected about you during the course of the research will be kept strictly confidential. All survey responses are anonymous, so it will not be possible to identify you in any reports or publications.

10. What will happen to the results of the research project?
The results of the project will be written up in a report for the funder (see below), and will be published in academic journals and presented at conferences. You will not be identified in any reports or publications. The anonymised data collected during the course of the project may be used for additional or subsequent research and analysis.

11. Who is organising and funding the research?
The project is being organised by Koonal Shah and Professors Allan Wailoo and Aki Tsuchiya of the University of Sheffield's School of Health and Related Research. It has been funded by NICE through its Decision Support Unit.

12. Who has ethically reviewed the project?
The study has been reviewed and approved by the Research Ethics Committee at the School of Health and Related Research (ScHARR) via the University of Sheffield Ethics Review Procedure.

13. Contact for further information
For information regarding participant recruitment, or if you no longer wish to participate, please contact Koonal Shah (k.k.shah@sheffield.ac.uk; 020 7747 8856). For any other issues, please contact Professor Aki Tsuchiya (a.tsuchiya@sheffield.ac.uk; 0114 222 0710).

At the end of the survey, we will provide details of resources for participants who are affected by or wish to seek further information about the topics covered in this project.

Thank you for reading this information sheet.

If you agree to take part, please complete the informed consent form.

Continue →

Appendix 18 Empirical study 4 pilot interview guide

Health Priorities Survey – Questions for participants

ID:

General feedback on survey

("I will begin with some general questions about the survey.")

1. How did you find the survey?

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

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

Feedback on scenarios / choice tasks

("I will now ask you about the hypothetical scenarios, in which you were asked to choose which patient the health service should treat.")

4. <Show wording of indifference option> What did you think about the option that did not involve choosing to treat either patient A or patient B?

5. <Show example diagram> What did you think about the diagrams used to illustrate the scenarios?

6. <Show example second question> 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?

7. <Show example second question> You were then asked to select your response from a list. Did the response you had in mind appear in this list?

8. <Show S3> This scenario 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?

9. <Show S6> This scenario 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?

10. <Show S8> This scenario 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?

Feedback on attitudinal questions

("I will now ask about the questions that followed the hypothetical scenarios.")

11. <Show attitudinal questions> These questions asked you to indicate the extent to which you agreed or disagreed with a particular statement. What did you think of these questions?

General feedback on survey

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

Appendix 19 Empirical study 4 ethics approval

Downloaded: 22/02/2017
Approved: 04/01/2016

Koonal Shah
Registration number: 100137338
School of Health and Related Research
Programme: ScHARR PhD PT

Dear Koonal

PROJECT TITLE: Valuing health at the end of life: methodological study (lay title: Health priorities survey)
APPLICATION: Reference Number 006867

On behalf of the University ethics reviewers who reviewed your project, I am pleased to inform you that on 04/01/2016 the above-named project was **approved** on ethics grounds, on the basis that you will adhere to the following documentation that you submitted for ethics review:

- University research ethics application form 006867 (dated 04/01/2016).
- Participant information sheet 1013708 version 2 (26/11/2015).
- Participant information sheet 1014582 version 1 (04/01/2016).
- Participant information sheet 1014581 version 1 (04/01/2016).
- Participant consent form 1013709 version 1 (24/11/2015).
- Participant consent form 1014584 version 1 (04/01/2016).
- Participant consent form 1014583 version 1 (04/01/2016).

If during the course of the project you need to deviate significantly from the above-approved documentation please inform me since written approval will be required.

Yours sincerely

Jennifer Burr
Ethics Administrator
School of Health and Related Research

Appendix 20 Empirical study 4 follow-up question responses

In the following charts (Figure 0-1 to Figure 0-10), each bar refers to the proportion of all respondents in the relevant arm (excluding those who expressed indifference in their initial choice, and therefore did not proceed to the follow-up question) who selected the specified value as their point of indifference. The graphs for S4, S5 and S6 are split between respondents who initially chose the life extension option and respondents who initially chose the quality of life improvement option.

Figure 0-1. S1 follow-up question responses (initial choice: 12-mth life extension) – n=1,770

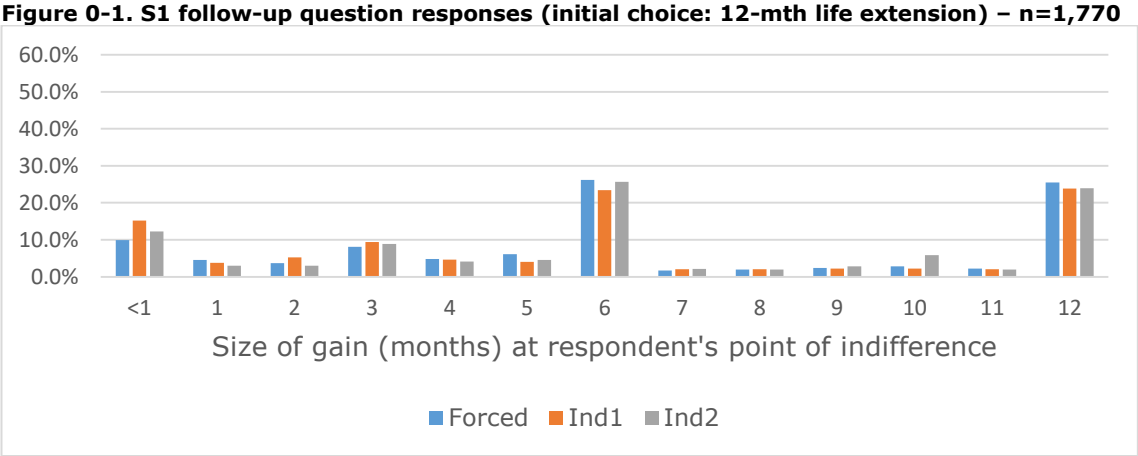


Figure 0-2. S2 follow-up question responses (initial choice: 12-mth life extension) – n=1,984

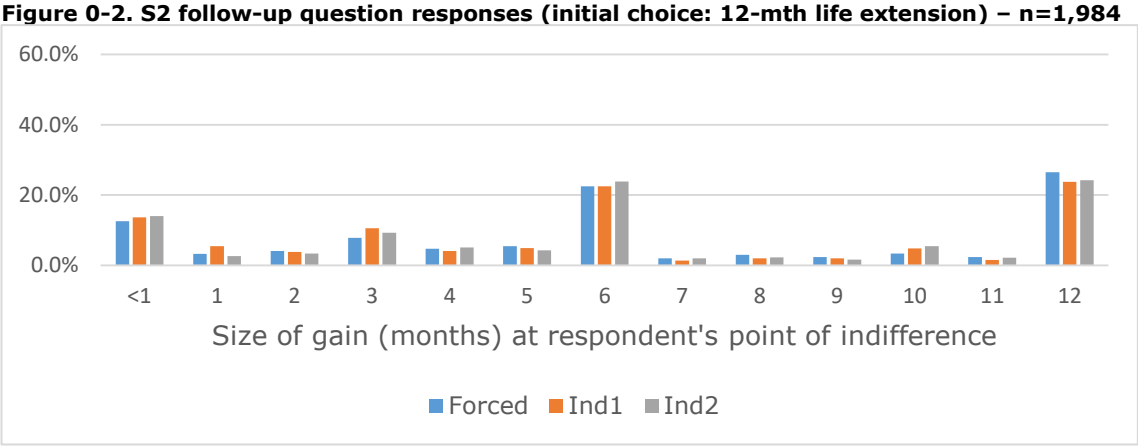


Figure 0-3. S3 follow-up question responses (initial choice: 12-mth life extension) – n=1,676

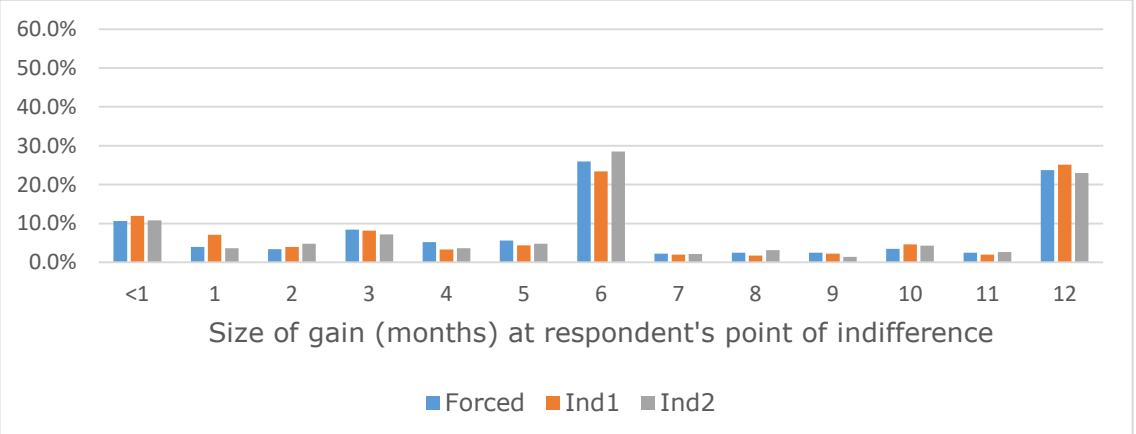


Figure 0-4. S4 follow-up question responses (initial choice: 12-mth life extension) – n=685

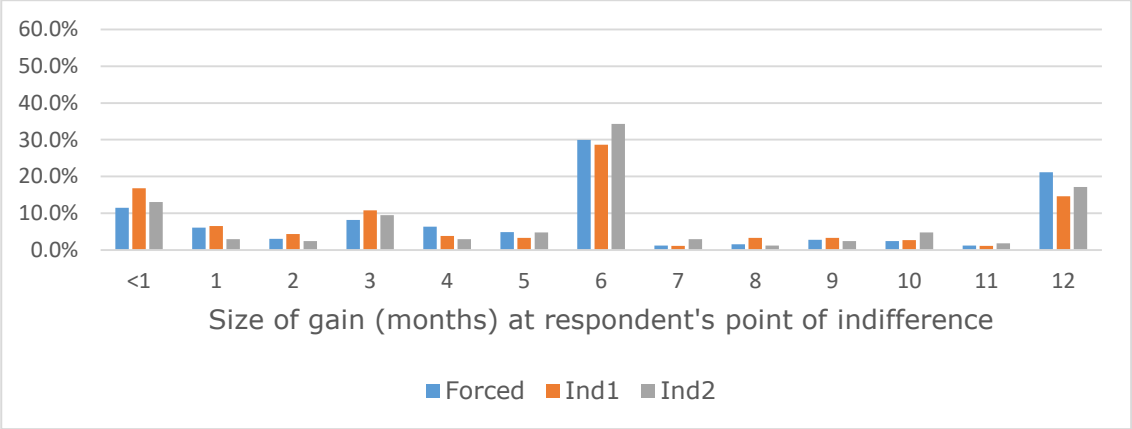


Figure 0-5. S4 follow-up question responses (initial choice: 50% QOL improvement) – n=1,094

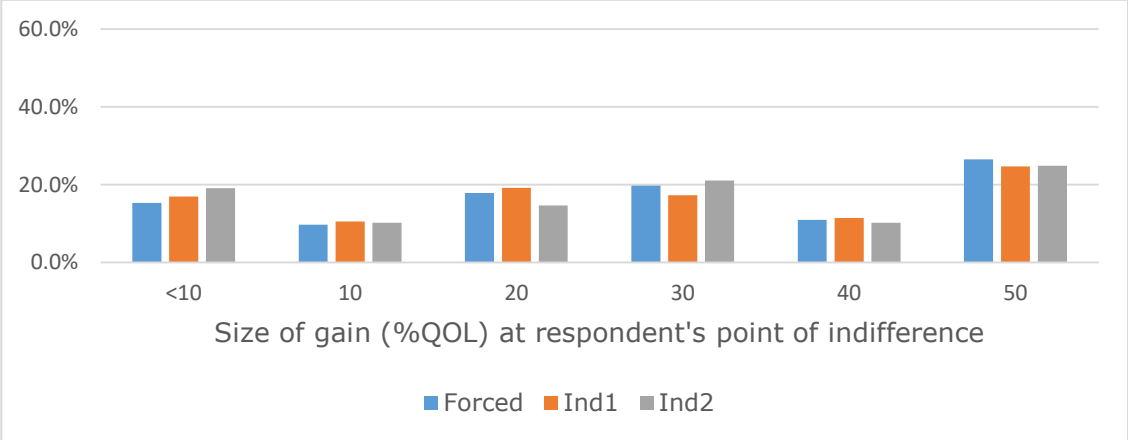


Figure 0-6. S5 follow-up question responses (initial choice: 12-mth life extension) – n=852

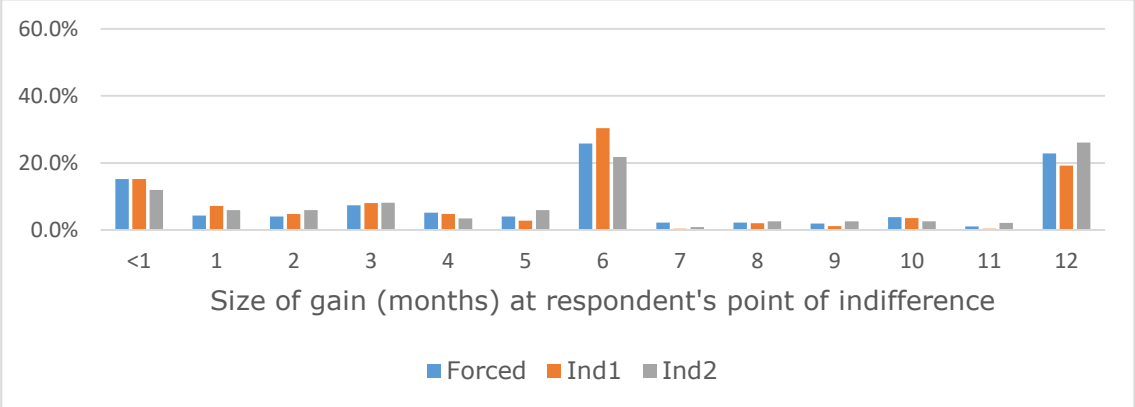


Figure 0-7. S5 follow-up question responses (initial choice: 50% QOL improvement) – n=1,030

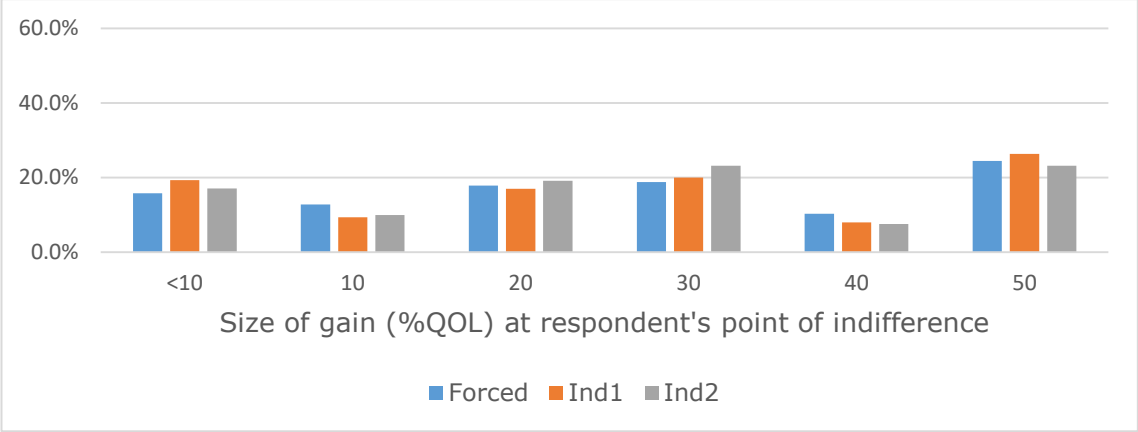


Figure 0-8. S6 follow-up question responses (initial choice: 12-mth life extension) – n=545

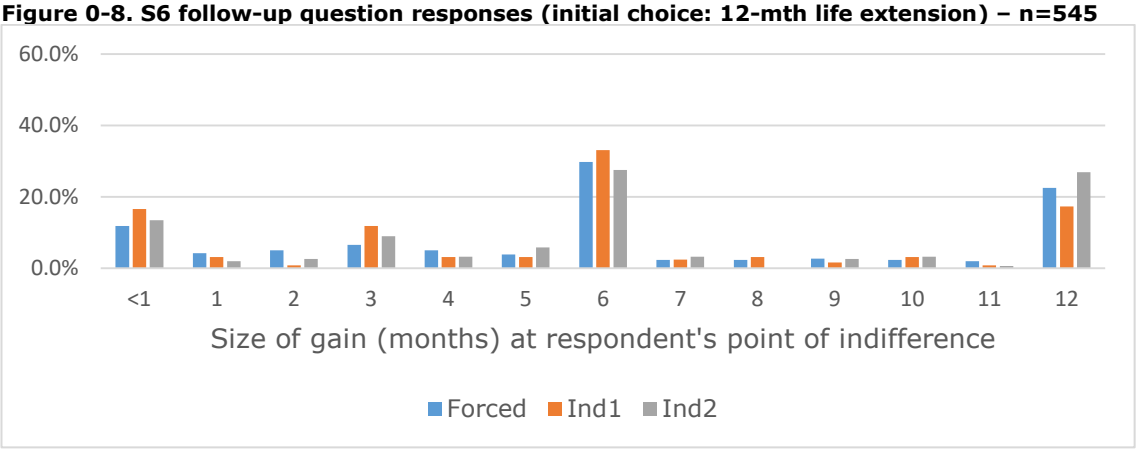


Figure 0-9. S6 follow-up question responses (initial choice: 50% QOL improvement) – n=1,167

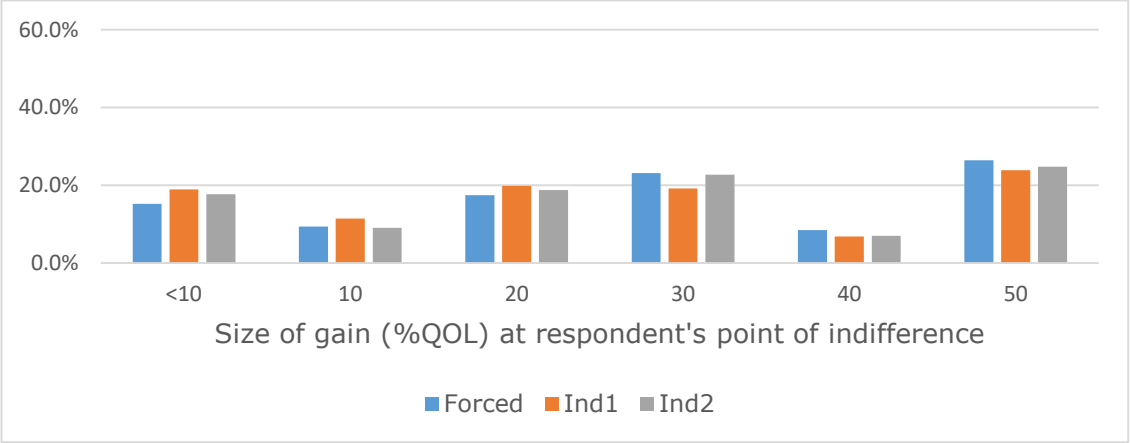
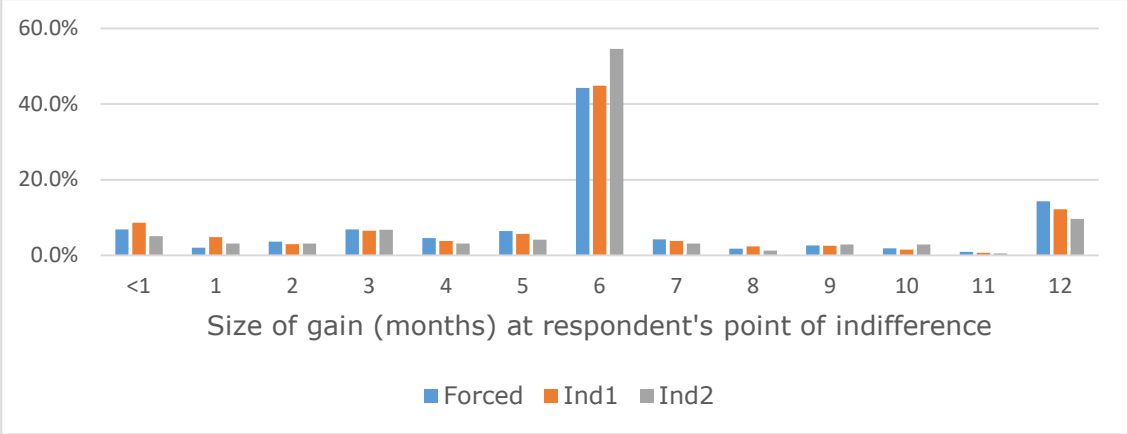


Figure 0-10. S7 follow-up question responses (initial choice: 6- or 12- mth life extension) – n=1,700



Appendix 21 Tabular comparison of empirical studies 1, 2, 3 and 4

Note: The fields in this table are the same as those in Table 2-1.

Study	**Empirical study 1**	**Empirical study 2**	**Empirical study 3**	**Empirical study 4**
Year of publication	Not fully published	2014	2015	Not fully published
Year of study conduct	2011	2011	2012	2016
Country or countries of origin of data	UK (England)	UK (England)	UK (England and Wales)	UK
Sample size	21	50	3,969	2,401
Type of sample	University staff and students	Public	Public	Public
Sample recruitment process	Flyers and email invitations	Door-knock	Internet panel	Internet panel
Criteria for excluding respondents and/or observations reported?	No	No	Yes – excluded respondents who spent insufficient time completing the survey	Yes – excluded respondents who spent insufficient time completing the survey
Mode of administration	Non-computer-assisted personal interview	Non-computer-assisted personal interview	Internet survey	Internet survey
Summary of primary study objective(s)	To pilot an approach for testing for support for end of life prioritisation	To test for support for end of life prioritisation	To test for support for end of life prioritisation	To test for framing effects in the elicitation of preferences regarding end of life
Was end of life (or a related term) mentioned explicitly in the study objectives?	Yes	Yes	Yes	Yes

Study	Empirical study 1	Empirical study 2	Empirical study 3	Empirical study 4
Pilot reported?	No (the study itself was a pilot)	Yes	Yes	Yes
Preference elicitation technique	Other choice exercise	Other choice exercise	Discrete choice experiment	Other choice exercise
Perspective	Social decision-maker	Social decision-maker	Social decision-maker	Both – study included one task that was an own health perspective operationalisation of an earlier social decision-maker perspective task
End of life definition	Life expectancy without treatment	Life expectancy without treatment	Life expectancy without treatment; dummy variable in the modelling representing the cut-offs associated with NICE criteria	Life expectancy without treatment
Life expectancy without treatment attribute levels	1yr, 10yrs	1yr, 10yrs	3mths, 12mths, 24mths, 36mths, 60mths	1yr, 5yrs, 30yrs
Life expectancy gain from treatment attribute levels	6mths, 1yr	6mths, 1yr	0mths, 1mth, 2mths, 3mths, 6mths, 12mths	6mths, 1yr
Was disease labelled or named?	No	No	No	No
Did the study examine whether quality of life-improving or life-extending treatments are preferred for end of life patients?	Yes	Yes	Yes	Yes
What were respondents	Which of two patients to treat	Which of two patients to treat	Which of two patients to treat	Which of two patients to treat

Study	Empirical study 1	Empirical study 2	Empirical study 3	Empirical study 4
choosing between (or choosing to do)?				
Was it possible to express indifference?	Yes – an 'I have no preference' option was available (but not listed with same prominence as other response options)	Yes – an 'I have no preference' option was available	No	Yes (in four of six arms; n=1,594); no (in two of six arms; n=807; for these respondents indifference could be expressed indirectly in a follow-up question)
Were visual aids used?	Yes	Yes	Yes	Yes (in three of six arms; n=1,202); no (in three of six arms; n=1,199)
Strength of preference examined at the individual respondent level?	No	No	No	Yes – using benefit trade-off type approach
Number of tasks completed by each respondent	5	6	10 (+2 further tasks examining the issue of preparedness)	8 (+attitudinal questions)
Time taken to complete survey reported?	Yes – between 20 and 35 minutes	No	No	Yes – 17 minutes on average
Summary of finding: end of life vs. non-end of life	Evidence consistent with an end of life premium	Evidence consistent with an end of life premium	Evidence not consistent with an end of life premium	Evidence not consistent with an end of life premium
Summary of finding: quality of life improvement vs. life extension	Quality of life improvement preferred	Quality of life improvement preferred	Life extension preferred	Quality of life improvement preferred
Other results of potential interest	Various insights into how design of an end of life preference study can be improved – e.g. explicit	No evidence that age- or time-related preferences are motivating factors for choosing to	Majority of respondents supported a mixture of the QALY-maximisation and priority-to-	Respondents' choices found to be sensitive to the inclusion of indifference options and the use

Study	Empirical study 1	Empirical study 2	Empirical study 3	Empirical study 4
	inclusion of an indifference option; some respondents reported that their views may be influenced by the life stages of the patients	treat end of life patient; no evidence that concern about the life stage of end of life patients is a motivating factor for preferring either life-extending or quality of life-improving treatments for those patients	worst-off approaches to priority-setting	of alternative study perspectives; preference for treating the end of life patient weakened when that patient is older than and/or has known about their prognosis for longer than the non-end of life patient
Other factors examined	Age, time preference	Age, time preference	Quality of life without treatment, preparedness	Age, preparedness, perspective, framing effects
Impact of background characteristics reported?	No	No	Yes – no characteristics found to be associated with preferences	Yes – respondents who are younger, have children and have experience of terminal illness were more likely to favour treating the end of life patient
Were qualitative data or explanatory factors sought?	Yes – respondents answered probing debrief questions as part of interview	Yes – respondents indicated the reasons for their choices by choosing from a list	No / not reported	Yes – via attitudinal questions
Was any reference made to age-related preferences?	Yes – no evidence that age is a motivating factor for choosing to treat end of life patient	Yes – no evidence that age is a motivating factor for choosing to treat end of life patient	No – age attribute was purposely omitted	Yes – evidence that age is a motivating factor for choosing to treat end of life patient
Was any reference made to time-related preferences?	Yes – one task involved choosing between a patient who had known their prognosis for some time and another who had only just learned their prognosis (life expectancy without treatment was the same for both)	Yes – one task involved choosing between a patient who had known their prognosis for some time and another who had only just learned their prognosis (life expectancy without treatment was the same for both)	Yes – two tasks involved choosing between a patient who had known their prognosis for some time and another who had only just learned their prognosis	Yes – one task involved choosing between a patient who had known their prognosis for some time and another who had only just learned their prognosis

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