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Attributes and weights in health care priority setting: a systematic review of what counts and to what extent

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Abstract

In most societies resources are insufficient to provide everyone with all the health care they want. In practice, this means that some people are given priority over others. On what basis should priority be given? In this paper we are interested in the general public's views on this question. We set out to synthesis what the literature has found as a whole regarding which attributes or factors the general public think should count in priority setting and what weight they should receive. A systematic review was undertaken (in August 2014) to address these questions based on empirical studies that elicited stated preferences from the general public. Sixty four studies, applying eight methods, spanning five continents met the inclusion criteria. Discrete Choice Experiment (DCE) and Person Trade-off (PTO) were the most popular standard methods for preference elicitation, but only 34% of all studies calculated distributional weights, mainly using PTO. While there is heterogeneity, results suggest the young are favoured over the old, the more severely ill are favoured over the less severely ill, and people with self-induced illness or high socioeconomic status tend to receive lower priority. In those studies that considered health gain, larger gain is universally preferred, but at a diminishing rate. Evidence from the small number of studies that explored preferences over different components of health gain suggests life extension is favoured over quality of life enhancement; however this may be reversed at the end of life. The majority of studies that investigated end of life care found weak/no support for providing a premium for such care. The review highlights considerable heterogeneity in both methods and results. Further methodological work is needed to achieve the goal of deriving robust distributional weights for use in health care priority setting.

Keywords

Health care priority setting; Stated preferences; Distributional weights; Relative weights; DCE; PTO; Systematic review

1. Introduction

Countries around the world face the question of how best to set priorities in the allocation of scarce health resources. Traditionally, economic evaluation has been adopted as the preferred approach to guide policy making in such decisions, with the quality-adjusted life year (QALY), a metric of health gain that combines both quality of life and length of life, being the most-commonly adopted measure of the value of a health care treatment (Drummond, 2013; Hjelmgren, et al., 2001). However, evidence from studies involving members of the public in various countries (Stafinski, et al., 2011; Whitty, et al., 2014a), from studies of past health technology assessment (HTA) decisions (Clement, et al., 2009; Devlin and Parkin, 2004; Harris, et al., 2008) and from HTA guidelines (Canadian Agency for Drugs Technologies in Health, 2006; National Institute for Health and Clinical Excellence, 2008; Pharmaceutical Benefits Advisory Committee, 2013) suggests that QALY gain may not be the sole determinant of value.

A key question is therefore what additional factors the general public (whose taxes contribute to funding health care) find important in resource allocation decisions? A second question is what distributional (or relative) weights these factors should receive in priority setting. Such weights attach different (numerical) importance to QALYs based on the characteristics of the beneficiaries.

These two questions have been explored extensively in the health economics research literature (the first more so than the second). However, little is known about what the literature, as a whole, has found regarding which factors or attributes the general public think should count, and to what extent, in health resource allocation decisions.

This systematic review focuses on studies that have elicited stated preferences from the public as to what factors should count in allocating publicly-funded health care; and the weights to be attached to such factors. We address four questions: (1) which methods have been used to elicit

stated preferences for attributes considered important in priority setting?; (2) which attributes arise out of the application of such methods?; (3) which methods have been used to elicit distributional weights?; and (4) what are the estimated distributional weights?

A small number of literature reviews on preferences in relation to priority setting have been conducted (Dolan, et al., 2005; Schwappach, 2002a; Shah, 2009; Stafinski, et al., 2011; Whitty, et al., 2014a; Youngkong, et al., 2009). In general they addressed our second question (an exception is Whitty, et al. (2014a) which also addressed our first question). However, to understand the differences between results about preferences it is necessary to understand how they were generated. Of importance too, after a large number of research studies, is whether a dominant approach has emerged for elicitation of preferences and weights, and whether a set of weights is ready for use in priority setting.

Thus, this paper contributes to the literature by, first, addressing new and important questions and, second, more completely addressing questions previously considered. In contrast to previous reviews, we consider a broader range of attributes and summarise them under three categories: (a) the characteristics of beneficiaries of the health gain/health care; (b) the characteristics of health gain; and (c) other important contextual factors. We highlight the degree of consensus amongst the literature and, wherever possible, identify possible reasons for differences. We focus on studies using samples representing the general public based on age and gender. Our review covers a broader range of methods and, importantly, compares results from different methods. It is also the first to classify the perspectives used in each study based on the six categories proposed by Dolan, et al. (2003).

2. Methods

2.1. Overview

PRISMA guidelines were used for the design of the review. The quality of studies included in the review was not evaluated because there is no single approach to assess the quality of the studies across the variety of methods used.

2.2. Literature search

Four databases were searched to ensure coverage across medicine and economics: Ovid Medline (1946 to present with daily update), Embase, Econlit and Web of Science (SCI-expanded and SSCI) from conception to 13 August 2014. Search terms were developed for two categories: health care priority setting (defined as setting priorities in the context of the allocation of publicly-funded health care) and preferences and were initially developed for the Ovid Medline database then modified for each database. Studies not reported in English were excluded from the review during screening and eligibility assessment. The complete search strategy, including search terms, for all four databases is in *Supplementary Appendix 1*. Additional studies were identified via a hand search of the references and citations from the included articles.

2.3. Eligibility criteria

Inclusion/exclusion criteria are in Table 1. Further to above, included studies were peer reviewed and must have involved elicitation of stated preferences between competing criteria, interventions, or patient groups applicable to priority setting for publicly-funded health care. Studies should not be disease or treatment specific in order to identify generic attributes, although some included studies provided clinical information merely as part of the question framing. Studies were excluded if their sample came from subgroups of the general public due to the focus on studies using samples representative of the general public, particularly in terms

of age and gender. Studies using students do not provide such representativeness while those using small samples do. The inclusion/exclusion criteria were sequentially applied, starting with the language requirement and ending with the sample criterion.

[INSERT TABLE 1]

Two authors (YG and PG) independently screened the titles and abstracts of all studies identified from the search strategies. Full-texts of the studies included after the initial screening were also independently reviewed by these two authors. Discrepancies were resolved by discussion until a consensus was reached.

2.4. Information extraction

Data were extracted based on the four questions outlined in the introduction and also according to: (a) characteristics of beneficiaries of the health gain/health care; (b) characteristics of health gain; and (c) other important contextual factors. We note that classification into these three groups is subjective and other classifications are possible.

3. Results

3.1. Overview

The process of study selection is summarised in Figure 1. The database and hand searches initially identified 4,504 studies, with 64 papers included in the review. Table 2 documents the complete list of studies (and attributes explored in each). Their detailed summary are in *Supplementary Appendix 2*.

[INSERT FIGURE 1]

[INSERT TABLE 2]

The 64 studies spanned over two decades from 1989 to 2014, Figure 2 revealing an increasing trend during this period. This may also reflect that earlier studies were excluded because they used non-representative samples of the general public. Studies were mostly carried out in the UK (n=20), the US (n=11), and Australia (n=9). Sample sizes varied from 23 to 17,657 with a median of 556. Around 70% of studies used a sample size larger than 200, and 55% greater than 500 (See *Supplementary Appendix 2*).

[INSERT FIGURE 2]

3.2. Methods for stated preference elicitation

Eight preference elicitation methods have been used, which we categorise as choice based techniques or ranking based techniques (see Figure 3). Choice based approaches have dominated the literature (used 62 times in 60 of the 64 studies) with ranking tasks used in 4 studies. Within choice based approaches, DCE (including one best worst scaling study) has been used 17 times, PTO 15 times, choice tasks with bespoke design 15 times, simple choice tasks varying a single attribute 9 times, contingent valuation method (CVM) (or Willingness to Pay (WTP)) 5 times and a choice experiment with allocation of points once. The popularity of DCE has been increasing over time – 50% of studies using DCE have been conducted in the past five years while only a third of the total PTO studies were undertaken during that time.

[INSERT FIGURE 3]

Following Dolan, et al. (2003), the perspective framing used in the included studies was classified based on (1) whom the respondent was asked to think about (personal, impartial decision maker (excluding self), citizen (including self)) and (2) the point in time at which the preference was elicited (ex post, ex ante). Most studies did not explicitly state the perspective used and thus classification was largely based on our interpretation. We identified 66

perspectives from 63 studies (three studies included more than one perspective). One study did not provide any information about the question context so could not be classified (Lim, et al., 2012). Thirteen of 65 perspectives were classified as impartial decision maker, three as citizen and four as personal. The remaining 46 could be either impartial decision maker or citizen depending on how respondents interpreted the context – we classified this as (non-specific) social perspective. The most commonly used timing perspective was ex post (58 times), followed by ex ante (7 times). One study could not be classified due to inconsistent statements within the question context (Mortimer and Segal, 2008).

3.3. Attributes and stated preferences

In eliciting preferences, most studies attempted to control for confounding factors, either via wordings such as “all else being equal” or via explicitly fixing factors such as health gain and the cost of treatment. DCE studies typically included the health gain as an attribute.

3.3.1. Characteristics of the beneficiaries

Age

Twenty-five studies (39%) elicited preferences for age. The majority (14 studies) suggest that the public in general favours the young over the elderly. However, preferences for age can be confounded by preferences for remaining life expectancy from a given age. If the confounding effect is controlled for, then the preference can be interpreted as preference for age per se; otherwise the preference for age may be conflated with preferences for remaining life expectancy. Among the 14 studies, eight controlled for the confounding effect (Dolan and Tsuchiya, 2005; Dolan and Tsuchiya, 2012; Johri, et al., 2009; Lewis and Charny, 1989; Nord, et al., 1996; Olsen, 2013; Schwappach and Strasmann, 2006; Tsuchiya, et al., 2003) while the

remaining six did not. In the latter case, the young being favoured over the elderly might simply be a reflection of the general public's preference for a person's capacity to benefit.

Another eight studies suggest that preferences for age display an inverted U pattern, implying a person at working age is valued most (Baker, et al., 2010; Charny, et al., 1989; Cropper, et al., 1994; Diederich, et al., 2012; Jelsma, et al., 2002; Palanca-Tan, 2013; Petrou, et al., 2013; Whitty, et al., 2014b). Of these, five controlled for the confounding effect of remaining life expectancy (Baker, et al., 2010; Charny, et al., 1989; Jelsma, et al., 2002; Palanca-Tan, 2013; Petrou, et al., 2013) while the other three did not.

Discordant evidence comes from the remaining three studies. Diederich, et al. (2011) reported that age had little effect on health care priority setting while Linley and Hughes (2013) suggested that children were not favoured over adults. Both studies provided the option of equal allocation to different populations which was chosen by the majority of respondents. Lancsar, et al. (2011) found that age at onset and age at death if untreated had little effect and a very small effect respectively. This could be due to the fact that, unlike the majority of studies in the review, respondents were able to trade-off age against the size of the health gain and the resulting preference for the size of the health gain outweighed the preference for age. Both Linley and Hughes (2013) and Lancsar, et al. (2011) controlled for the confounding effect of remaining life expectancy while Diederich, et al. (2011) did not.

In total 15 studies controlled for the confounding effect of remaining life expectancy through four approaches: (1) four studies explicitly assumed the remaining life expectancy from current age is the same (Dolan and Tsuchiya, 2005; Johri, et al., 2009; Nord, et al., 1996; Olsen, 2013); (2) four studies used wordings such as "all else being equal" (Charny, et al., 1989; Lewis and Charny, 1989; Linley and Hughes, 2013; Palanca-Tan, 2013); (3) two considered age and the remaining life expectancy simultaneously (Lancsar, et al., 2011; Schwappach and Strasmann,

2006); while (4) five studies explicitly stated in the survey that the preference is about a fixed number of years of life in full health (or a given quality of life (QoL)) at different ages (Baker, et al., 2010; Dolan and Tsuchiya, 2012; Jelsma, et al., 2002; Petrou, et al., 2013; Tsuchiya, et al., 2003). The remaining 10 studies did not control for the confounding effect of remaining life expectancy. Whether the preferences elicited in these studies are about age per se or about remaining life expectancy will depend on how respondents interpreted the question context and the assumptions they made..

Severity

Severity is included as a characteristic of the beneficiary since severity of an illness is experienced by the patient. Nineteen studies (30%) elicited preferences for severity. There is considerable heterogeneity in the definitions of severity used in the literature. Table 3 documents the use of three types of severity related attributes. Not all 19 studies explicitly defined these attributes as severity but all three types have been explicitly defined as severity in the literature. The most popular definition is based on ‘QoL if untreated’. Four studies also defined severity in terms of life expectancy (LE) if untreated (or age of onset and age of death if untreated) (Dolan and Tsuchiya, 2005; Lancsar, et al., 2011; Lim, et al., 2012; Whitty, et al., 2011). A number of studies loosely referred to pre-treatment health problems but did not describe the specific problems (Dolan and Shaw, 2003; Green, 2009; Green and Gerard, 2009; Linley and Hughes, 2013; Ryyanen, et al., 1996; Ubel, 1999; Winkelhage and Diederich, 2012).

[INSERT TABLE 3]

Nevertheless, the empirical evidence is consistent with 19 (out of 22) studies suggesting that members of the general public are in general willing to give priority to a patient with more

severe disease. Among these studies, three further highlight that severity may be one of the most important attributes to use in health care priority setting: Dolan and Shaw (2003) and Diederich, et al. (2012) both ranked severity as the most important attribute and Linley and Hughes (2013) reported that ‘more severely ill’ was favoured regardless of size of health gain or cost of treatment.

Discordant evidence comes from the remaining three studies. Dolan and Tsuchiya (2005) and Lancsar, et al. (2011) found small or no effects of severity on priority setting with the small effect favouring the less severely ill. In the former, preference for severity is outweighed by the preference for the size of the health gain; in the latter respondents may be quite divided between favouring severe and less severe conditions, along with indifferent respondents. Ubel (1999) found that preferences for prioritising more severely ill patients were sensitive to the question framing.

Lifestyle/Self-induced illness

Ten studies (16%) elicited preferences for lifestyle or self-induced illness. All suggest that the general public would give less priority to those considered in some way responsible for their ill health. However, Edlin, et al. (2012) found that this preference against self-induced illness was outweighed by the preference for prioritising the more severely ill.

Socioeconomic status (SES)

Ten studies (16%) elicited preferences relating to beneficiaries’ SES. Eight suggest that respondents tend to favour individuals with low SES over those with high SES. The other two suggest that people’s SES had little effect on priority setting (Charny, et al., 1989; Linley and Hughes, 2013).

Other attributes of beneficiaries

Other attributes were explored by a relatively small number of studies, including having dependents, marital status, employment status, gender, indigenous people, QoL after treatment, treatment potential, rarity of disease, lifetime QALYs (i.e., health from a specific time to death), past QALYs (i.e., health experienced up to now), and past QoL (i.e., QoL of all the past years). Results are summarised in *Supplementary Appendix 3*.

3.3.2. Characteristics of health gain

Size/distribution of the health gain

Twenty three studies (36%) elicited preferences for size or distribution of the health gain. Again there is heterogeneity in the definition of health gain used in the literature including: gain in life years; QoL; and QALYs. If QoL is fixed, gain in life years would imply gain in QALYs. However, when using gain in life years, studies in general did not assume QoL to be fixed. Larger gains are universally preferred over smaller gains and size of health gain was considered one of the most important attributes for health care priority setting in 3 studies (Dolan and Shaw, 2003; Lancsar, et al., 2011; Linley and Hughes, 2013).

Four studies suggest that the preference for larger gains may diminish as the size of gain increases (Lancsar, et al., 2011; Nord, et al., 1996; Norman, et al., 2013; Richardson, et al., 2011). This is in line with the finding of another six studies which suggest that people prefer to give small gains to many rather than large gains to a few (Gyrd-Hansen and Kristiansen, 2008; Olsen, 2000; Richardson, et al., 2012; Ubel, et al., 2001; Ubel, et al., 2000; Ubel, et al., 1996). However, Gyrd-Hansen and Kristiansen (2008) and Olsen (2000) found this preference for dispersion of gain over concentration happens only when the gain per person is greater than a certain threshold.

Prevention vs. cure

Five studies (8%) elicited preferences between prevention and cure. The majority suggest that prevention is preferred; the exception being Corso, et al. (2002) who found that cure was favoured over prevention based on median WTP estimates. During preference elicitation, all five studies kept the size of the health gain constant and all but Corso, et al. (2002) kept costs constant. However, WTP studies generally do not consider the costs of treatments. This may explain Corso, et al. (2002)'s unexpected result as some respondents may have thought cure is more costly than prevention and therefore were willing to pay more for an expensive good than for a cheap good.

Components of health gain

Five studies (8%) elicited preferences for different components of health gain. Three components have been identified from the literature including the gain from QoL enhancement, from life extension, and from lifesaving (prevention of premature death). Results overall generally suggest gain from life extension is favoured over the gain from QoL enhancement (Olsen and Donaldson, 1998; Pennington, et al., 2013; Whitty, et al., 2014b). However, this preference may not apply to all life stages, e.g., at the very end of life, the gain from QoL improvement becomes more valuable than the gain from life extension (Pinto-Prades, et al., 2014; Shah, et al., 2014). Finally, Olsen and Donaldson (1998) showed that a lifesaving gain was valued much more highly than the other two types of health gain. Among the five studies, all but Whitty, et al. (2014b) fixed the size of the health gain, but none fixed the costs of treatments. Again, three were WTP studies which in general do not consider costs of treatments.

Other health gain attributes

Two further attributes related to the characteristics of health gain have been explored: the timing of saving lives with Cropper, et al. (1994) finding people attach less importance to saving lives in the future than the present, and geographic equality of health gain distribution which was favoured by the respondents in a study by Quintal (2009).

3.3.3. Other important contextual factors

Cost of treatment

Eight studies (13%) elicited preferences for the cost of treatment. The majority (five studies) suggest a preference for lower cost. However, two studies found higher cost was preferred (Linley and Hughes, 2013; Ryyananen, et al., 1996) and one found cost had little effect (Nord, et al., 1995). Plausible explanations have been offered for this surprising result: Ryyananen, et al. (1996) stated that respondents potentially associated expensive care with severe disease while Linley and Hughes (2013) suspected that the elicited preference related to a general preference for fairness in receiving treatment based on need. All but Ryyananen, et al. (1996) and Nord, et al. (1995) fixed the size of the health gain.

Other contextual attributes

Several other contextual factors were explored by a small number of studies. These include availability of alternative treatment, number of patients, chance of success, cost effectiveness, and uncertainty in evidence. Results are included in *Supplementary Appendix 3*.

3.3.4. Cross-cutting attribute: end of life treatment

End of life treatment is defined by the National Institute for Health and Care Excellence (NICE) as a treatment that meets three criteria based on life expectancy if untreated (less than 24 months), life expectancy gain from treatment (at least 3 months), and the size of patient

population (small population) (National Institute for Health and Clinical Excellence, 2009). We single it out as a cross-cutting attribute since these criteria correspond to all three types of attributes: severity, size of health gain, and number of beneficiaries. Seven studies (11%) elicited preferences for end of life treatment which, in most cases, has been defined using only the first two criteria. Evidence is mixed: five studies provide weak or no support for favouring such treatment while the other two suggest moderate support (Pennington, et al., 2013; Pinto-Prades, et al., 2014).

3.4. Methods for elicitation of distributional weights

Among those using samples drawn from the general public, only 22 studies (34%) calculated distributional weights. Methods used to calculate relative weights included: PTO, DCE and social welfare function (SWF) based methods. Table 4 documents the 22 studies classified by methods and attributes.

[INSERT TABLE 4]

PTO was used in 15 studies. PTO asks respondents how many outcomes of one kind (condition A) are equivalent in social value to X outcomes of another kind (condition B). If one person's answer is Y, then this individual's weight between condition A and B are X/Y and 1. Typically individual responses have been aggregated in two ways: the ratio of means (or medians); or the mean (or median) of the ratios (Baker, et al., 2010).

Five studies used the SWF based approach. Typically a certain type of SWF needs to be chosen, and then its parameters are assumed and/or estimated through a bespoke choice exercise (a factorial experimental design is typically not involved except in Palanca-Tan (2013)) which usually involves multiple groups of patients at different levels of one attribute (e.g., age, severity, social class) and with different aggregate health. The distributional weights for the

attribute are estimated as the marginal rate of substitution between groups (Dolan and Tsuchiya, 2011).

Two studies derived weights using DCEs. Lancsar, et al. (2011) introduced a method based on the compensating variation to derive weights. In their study, two types of distributional weights were calculated: (1) weights for different levels of individual attributes and (2) weights for different combinations of attributes. Using the same DCE data, Baker, et al. (2010) also presented an approach called ‘predicted probability of choice’. It produced somewhat larger weights compared to the compensating variation based approach. Finally, Norman, et al. (2013) used the marginal rate of substitution, to calculate weights for different combinations of attributes.

3.5. Distributional weights

Most distributional weights were elicited for age (11/22 studies that elicited weights) and severity (4/22 studies that elicited weights). These results are discussed here. Results for attributes other than age and severity are documented in *Supplementary Appendix 2*.

Age

Since the methods, attribute levels and question framings vary from study to study, it is challenging to synthesise and compare the distributional weights estimated from these studies. Nevertheless, the smallest weights are reported from a study using a DCE. In Lancsar, et al. (2011), the weights for age of onset were all close to 1, implying no weighting. Such weights are calculated by changing age of onset from its reference level of 40 years, while holding all other variables at their reference level (QoL lost=0.7, age of death=60 and QALY gain=4). While the authors do not state this in their paper, given they control for the confounding effect of remaining life expectancy, this suggests the elicited weights can be interpreted as weights

for age per se. For age of death without treatment, higher weight was given to those who die young, aged 10, or die old, aged 70 or 80, although the weights remained relatively small. In contrast, the largest weights are reported from a study using PTO. In Johannesson and Johansson (1996), saving one person at age 30 was equivalent to saving 35 people at age 70, implying a weight of 35 to 1 between age 30 and 70. Apart from using different samples and sample sizes, there are two reasons that may explain this drastic difference. First, in Lancsar, et al. (2011) health gain is another attribute considered along with age and the analysis suggests that the choices were driven by the size of the health gain. Second, Johannesson and Johansson (1996) used a lifesaving context and did not explicitly fix the remaining life expectancy for patients of different ages. As previously discussed, respondents may conflate preferences for age per se and preferences for the remaining life expectancy in which case weights are more likely to represent the value of the remaining life expectancy from different ages rather than the value of one year in full health at different ages. Evidence suggests that the former may lead to larger weights favouring the young over the elderly than the latter (Jelsma, et al., 2002).

Severity

Compared to age weighting, an additional challenge to synthesise distributional weights for severity is that there is considerable heterogeneity in the definitions of severity used in the literature. Nevertheless, the smallest weights again come from a DCE study where significantly less weight (although still relatively close to 1) was given to the relatively more severe (Lancsar, et al., 2011). The largest weights are from a PTO study where saving one life was equivalent to returning 3 patients with severe health problems to full health (Nord, et al., 1993). The implied weights are 3 to 1 between saving a life and returning someone severely ill to full health.

4. Discussions and conclusions

This review has highlighted a large and increasing literature spanning five continents suggesting that the topic of preferences in relation to health care priority setting is highly relevant from an academic and policy perspective.

Empirical evidence has mainly focused on the characteristics of the recipients of health care and within this a particular focus on age and severity. Results generally suggest that the young are favoured over the old and the more severely ill are favoured over the less severely ill. There is evidence to suggest that people with self-induced illness or high SES should receive lower priority. Larger health gain is universally preferred, but potentially at a diminishing rate. These results are in general consistent with findings of past reviews. We also identified attributes that have previously been less explored. For example, a small number of studies suggest that life extension is favoured over QoL enhancement, although this preference may be reversed at the end of life. Also, our review suggests that, while in some jurisdictions higher weight is given to end of life treatment (National Institute for Health and Clinical Excellence, 2009), the majority of studies that investigated end of life care found weak/no support for providing a premium for such care.

This review is the first effort in the literature to review studies exploring distributional weights across a range of attributes. While 64 papers explored the broad topic of preferences for health care priority setting, only 34% calculated distributional weights. Where weights were derived they generally focused on age and severity, despite evidence in this review that other characteristics of recipients of the health gain may be important as may the characteristics of the health gain itself or other contextual factors. Deriving weights for a broader set of characteristics would be a natural extension warranting further research. The variation in the magnitude of the distributional weights reported in the literature is very large, likely due to

large heterogeneity in methods, contexts, countries/populations, sample sizes and the particular attributes (and their definitions) considered in the studies.

Eight different preference elicitation methods were used in the literature included in this review. There has been little consistency in methods although the majority of studies used a choice based approach with PTO and DCE being the most popular. Earlier in the literature PTO was the method most likely to be used but the more recent literature appears to be moving increasingly to using DCEs. DCEs have mainly been used to elicit preferences (usually investigated by examination of direction and significance of parameters estimated in the choice model). However, very few DCEs went on to derive distributional weights from the estimated choice models possibly because there has not been a standard method to derive weights. This would be a fruitful avenue for further research. In contrast, PTOs involve the direct elicitation of weights from which preferences are inferred. PTOs generally produced much larger distributional weights than DCEs. The focus in PTOs on single attributes one at a time rather than allowing trade-offs over multiple attributes could artificially inflate the resulting relative weights compared to DCEs which involve tradeoffs across multiple attributes simultaneously (Lancsar, et al., 2011). Similar results suggesting some strong isolated effects disappeared after the effects of additional attributes were taken into account were found by others using non-DCE and non-PTO methods (Edlin, et al., 2012; Linley and Hughes, 2013). However, this remains an unresolved issue. In light of the extreme differences in empirical results found from DCE and PTOs, sometimes even within the same study populations (Baker, et al., 2010), future work on theoretical and empirical reconciliation is important to ascertain the extent to which these differences are due to differences in preference elicitation methods and differences in actual preferences.

While using different methods may explain a large part of the variation in the estimated distributional weights, substantial differences also exist among studies using the same approach. One possible reason is that, while these studies derive weights for similar attributes, there is heterogeneity in the definitions of these attributes. In the case of age weighting, there is a distinction between value of a year of life at different ages and value of the remaining life from different ages. The latter may lead to larger weights between young and old. Similarly, severity is the attribute with the least consensus regarding its definition and description to respondents. Regarding health gain, despite the literature often being couched in terms of distributional weights for QALYs, few studies defined the health gain attribute in terms of QALYs. Future consistency in terms of attribute descriptions may be warranted but should be balanced against the need to define attributes such that they allow investigation of the research questions at hand within each study. Another possible reason for the wide variation in weights is that even within the same approach, the method used to calculate weights can differ. For example, within DCE, based on very limited evidence, it seems the compensating variation, predicted probability and the marginal rate of substitution approaches may lead to different results.

A number of areas would benefit from future research. One is interactions between attributes and their effect on preferences for priority setting and distributional weights. Given the complex and multifaceted nature of priority setting, it may be that preferences for one attribute are influenced by preferences for another. For example, does the size of the health gain affect preferences for prioritising across severity? Interactions were rarely considered in the literature covered in this review (with two exceptions: Whitty, et al. (2014b) and Norman, et al. (2013)) but are an important area warranting further research to uncover potentially important nuances in preferences and weights.

A large subset of papers included in the review loosely refer to social value and social preferences. While such studies generally did not provide explicit definitions (an exception is Gyrð-Hansen & Kristiansen, 2008), they loosely refer to social values or social preferences (and trade-offs between equity and efficiency) to distinguish such studies from individual preference based studies. When being asked to consider resource allocation within a publicly-funded system, studies that referred to social preferences and values generally asked respondents to think about community (with or without themselves being included in that group), whereas studies eliciting individual preferences involved respondents thinking only about themselves. It is not surprising that concepts of social value and preferences have not been well defined in the studies in this review given the debate in the literature more generally regarding social choice theory (Arrow, et al., 2010). However, future research in this area would benefit from defining such concepts more explicitly, at least in the context of study objectives and the interpretation of results.

A related issue is the perspective used for preference elicitation in the context of health care priority setting at a government level. Framing of perspectives used was generally poorly expressed and in some circumstances inconsistent with the choice context described to respondents. As for the personal, impartial decision making or citizen, ex post or ex ante, which to use will largely depend on a study's specific objective (Dolan, et al., 2003). Nevertheless, researchers are encouraged to distinguish these perspectives and to provide justification for their choices. Further research comparing the preferences and weights elicited from different perspectives is also warranted.

While important work has been undertaken to provide information on the general public's preferences in relation to health care priority setting, this review has highlighted a large degree of variation in both methods and empirical results. The literature seems to have converged

towards DCE as a dominant approach to deriving preferences for priority setting in this area. However, the calculation of weights within DCE is in its infancy but represents a fruitful area for further research. Within this and other methods, it is important to account for the methodological issues noted above. Such methodological work would assist in achieving the goal of deriving robust distributional weights that could assist policy makers faced with the important challenge of prioritising the allocation of scarce health care resources.

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Table 1 Inclusion and exclusion criteria for the literature review

	Inclusion criteria	Exclusion criteria
Language	- English	- Not in English
Publication	- Peer reviewed	- Not peer reviewed
Context	<ul style="list-style-type: none"> - Stated preferences - Health care setting - Include competing criteria, interventions, or patients in a general setting - Applicable to priority setting for publically-funded health care 	<ul style="list-style-type: none"> - Revealed preferences - Non health care setting - Criteria, interventions, or patients in a specific setting (e.g. HIV/AIDS, organ transplantation, informal/formal services) - Broad or narrow priority setting level (e.g. global priorities, priorities within a healthcare facility)
Data	- Empirical studies	- Other studies (e.g. theoretical studies, editorials, review articles)
Methods	- Quantitative methods for trade-offs (e.g. choice-based methods, person trade-off methods, and ranking or rating exercises).	- Qualitative methods (e.g. focus groups and structured interviews, percentage agreement with a statement).
Sample	- General public	<ul style="list-style-type: none"> - Subgroups of the general public (e.g. patients, clinicians, gender, specific age) - Health technology assessment decision makers

Table 2 Attributes explored in each study

Study	Age	Severity	Lifestyle/Self-induced illness	SES	Other attributes of beneficiaries	Size/distribution of health gain	Prevention vs cure	Components of health gain	Other health gain attributes	Cost of treatment	Other contextual attributes	End of life
Abasolo and Tsuchiya (2004)				✓								
Abásolo and Tsuchiya (2013)				✓								
Baker, et al. (2010)	✓	✓										
Bleichrodt, et al. (2005)					Lifetime QALYs							
Bosworth, et al. (2010)						✓	✓					
Bryan, et al. (2002)					QoL after treatment	✓					Number of patients, Chance of success	
Charny, et al. (1989)	✓		✓	✓	Marital status, Employment status, Gender							
Corso, et al. (2002)							✓					
Cropper, et al. (1994)	✓								Timing of saving lives			
Diederich, et al. (2012)	✓	✓	✓	✓	Having dependents, Marital status							
Diederich, et al. (2011)	✓											
Dolan and Shaw (2003)		✓	✓		Having dependents	✓						
Dolan and Tsuchiya (2005)	✓	✓			Past QoL							
Dolan and Tsuchiya (2009)			✓									
Dolan and Tsuchiya (2011)				✓								
Dolan and Tsuchiya (2012)	✓				Past QALYs							

Study	Age	Severity	Lifestyle/Self-induced illness	SES	Other attributes of beneficiaries	Size/distribution of health gain	Prevention vs cure	Components of health gain	Other health gain attributes	Cost of treatment	Other contextual attributes	End of life
Edlin, et al. (2012)			✓		Lifetime QALYs							
Eisenberg, et al. (2011)	✓											
Green (2009)		✓		✓								
Green and Gerard (2009)		✓				✓					Availability of alternative treatment, Cost effectiveness	
Gyrd-Hansen (2004)		✓										
Gyrd-Hansen and Kristiansen (2008)						✓						
Jelsma, et al. (2002)	✓											
Johannesson and Johansson (1996)	✓											
Johannesson and Johansson (1997)	✓											
Johri, et al. (2009)	✓											
Lancsar, et al. (2011)	✓	✓				✓						✓
Lewis and Charny (1989)	✓											
Lim, et al. (2012)		✓		✓		✓						
Linley and Hughes (2013)	✓	✓		✓	Having dependents, Rarity of disease	✓				✓	Availability of alternative treatment	✓
Mortimer and Segal (2008)	✓		✓			✓	✓			✓	Uncertainty in evidence	
Nord, et al. (1993)		✓										
Nord, et al. (1995)										✓		

Study	Age	Severity	Lifestyle/Self-induced illness	SES	Other attributes of beneficiaries	Size/distribution of health gain	Prevention vs cure	Components of health gain	Other health gain attributes	Cost of treatment	Other contextual attributes	End of life
Nord, et al. (1996)	✓					✓						
Norman, et al. (2013)			✓	✓	Having dependents	✓						
Olsen (2000)						✓						
Olsen (2013)	✓					✓						✓
Olsen and Donaldson (1998)								✓				
Palanca-Tan (2013)	✓					✓					Number of patients	
Pennington, et al. (2013)								✓				✓
Petrou, et al. (2013)	✓											
Pinto-Prades, et al. (2014)								✓				✓
Quintal (2009)									Geographic equality			
Richardson, et al. (2012)						✓						
Richardson, et al. (2011)		✓				✓						
Roberts, et al. (1999)					QoL after treatment							
Ryynanen, et al. (1996)	✓	✓	✓							✓		
Schwappach (2002b)							✓					
Schwappach and Strasmann (2006)	✓	✓			Rarity of disease	✓				✓		
Shah, et al. (2014)								✓				✓
Shiroiwa, et al. (2013)		✓										✓
Singh, et al. (2012)			✓									
Tsuchiya and Dolan (2007)				✓								

Study	Age	Severity	Lifestyle/Self-induced illness	SES	Other attributes of beneficiaries	Size/distribution of health gain	Prevention vs cure	Components of health gain	Other health gain attributes	Cost of treatment	Other contextual attributes	End of life
Tsuchiya, et al. (2003)	✓											
Ubel (1999)		✓										
Ubel, et al. (2001)						✓						
Ubel, et al. (2000)						✓						
Ubel, et al. (1996)						✓						
Ubel, et al. (1999)					Treatment potential							
Ubel, et al. (1997)		✓					✓					
Whitty, et al. (2014a)	✓				Indigenous people			✓			Availability of alternative treatment, Number of patients, Cost effectiveness	
Whitty, et al. (2008)					QoL after treatment	✓				✓	Chance of success	
Whitty, et al. (2011)		✓				✓				✓	Chance of success	
Winkelhage and Diederich (2012)	✓	✓	✓			✓				✓		

Note:

(1) SES: socioeconomic status

(2) QoL: Quality of life

Table 3 Descriptions of severity and frequency of use

Severity description	Frequency	Percentage
QoL ⁽¹⁾ if untreated or QoL loss if untreated (i.e., 1- QoL if untreated)	12	52
Pre-treatment health problems, not clearly defined	7	30
LE if untreated ⁽²⁾	4	17
Total	23	100

Note:

- (1) QoL means 'quality of life'.
- (2) LE means 'life expectancy'; Lancsar, et al. (2011) included LE implicitly via age of onset and age of death.
- (3) Several studies used more than one severity related attributes so the frequency of use here adds up to more than 19.

Table 4 Distributional weights classified by attributes and methods

Study	Age			Severity			Other attributes		
	PTO	DCE	SWF	PTO	DCE	SWF	PTO	DCE	SWF
Baker, et al. (2010)	✓	✓		✓	✓				
Cropper, et al. (1994)	✓								
Eisenberg, et al. (2011)	✓								
Jelsma, et al. (2002)	✓								
Johannesson and Johansson (1996)	✓								
Johannesson and Johansson (1997)	✓								
Nord, et al. (1993)				✓					
Nord, et al. (1996)	✓								
Petrou, et al. (2013)	✓								
Pinto-Prades, et al. (2014)							End of life		
Quintal (2009)							Geographic equality of health gain distribution		
Richardson, et al. (2011)				✓					
Schwappach (2002b)							Prevention vs cure		
Singh, et al. (2012)							Lifestyle		
Ubel, et al. (1999)							Treatment potential		
Bleichrodt, et al. (2005)									Lifetime QALYs
Dolan and Tsuchiya (2009)									Self-induced illness
Dolan and Tsuchiya (2011)									SES
Dolan and Tsuchiya (2012)			✓						Past QALYs
Palanca-Tan (2013)			✓						
Lancsar, et al. (2011)		✓			✓			Combination of several attributes including health gain	
Norman, et al. (2013)								Combination of several attributes including health gain	

Note: (1) PTO: Person trade-off ; (2) DCE: Discrete choice experiment; (3) SWF: Social welfare function; (4) SES: Socioeconomic status

Figure 1 Flow diagram for selection of studies

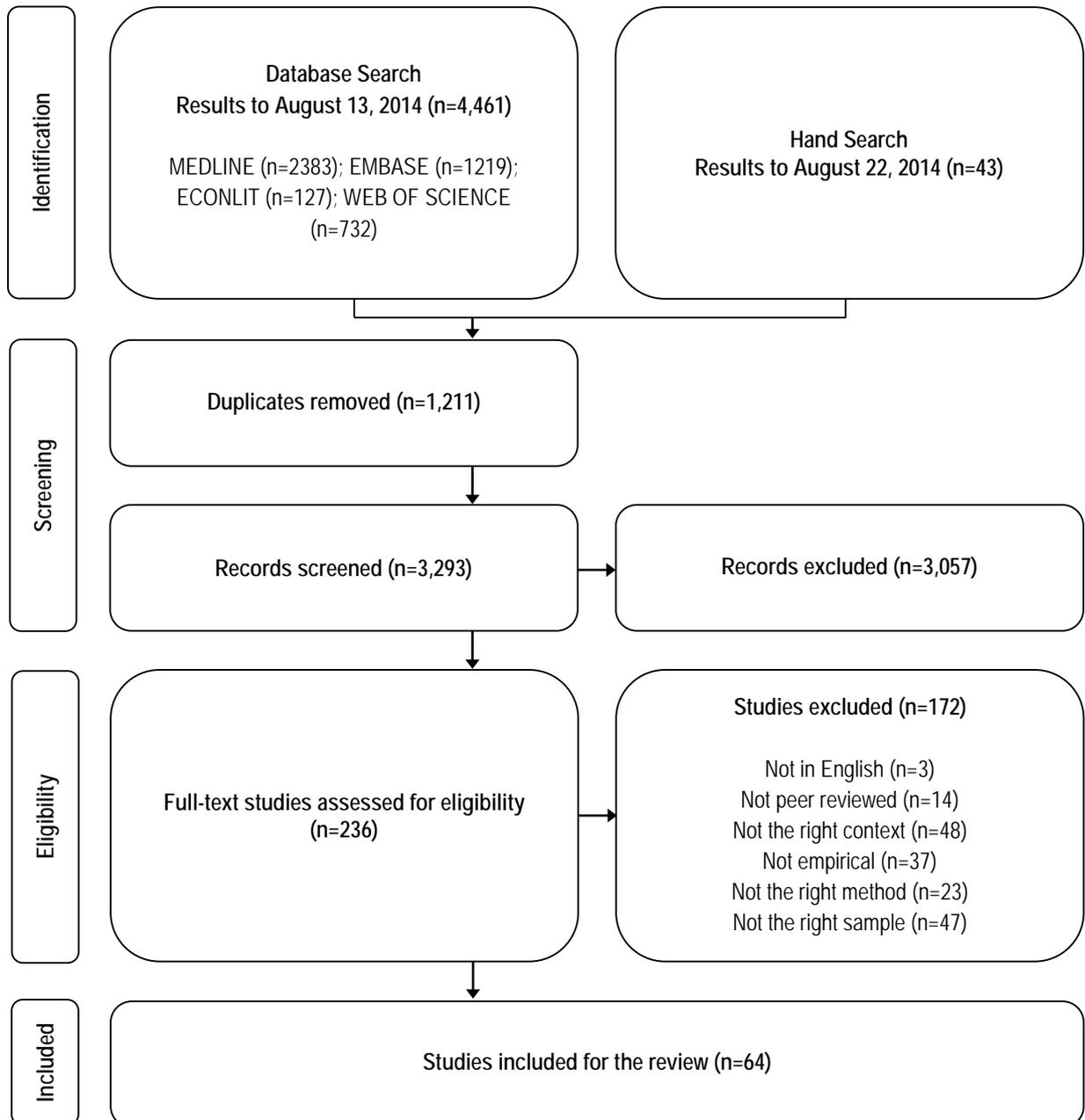


Figure 2 Number of studies per year from 1989 to August 2014

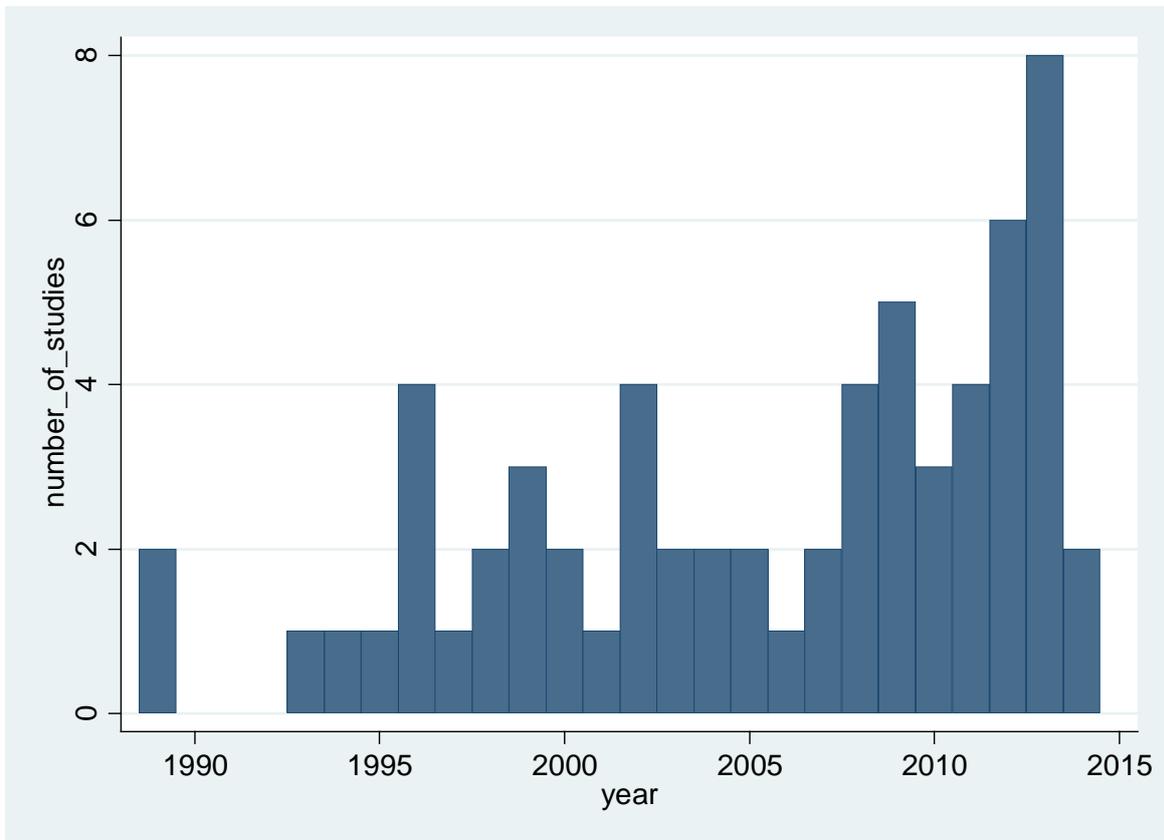
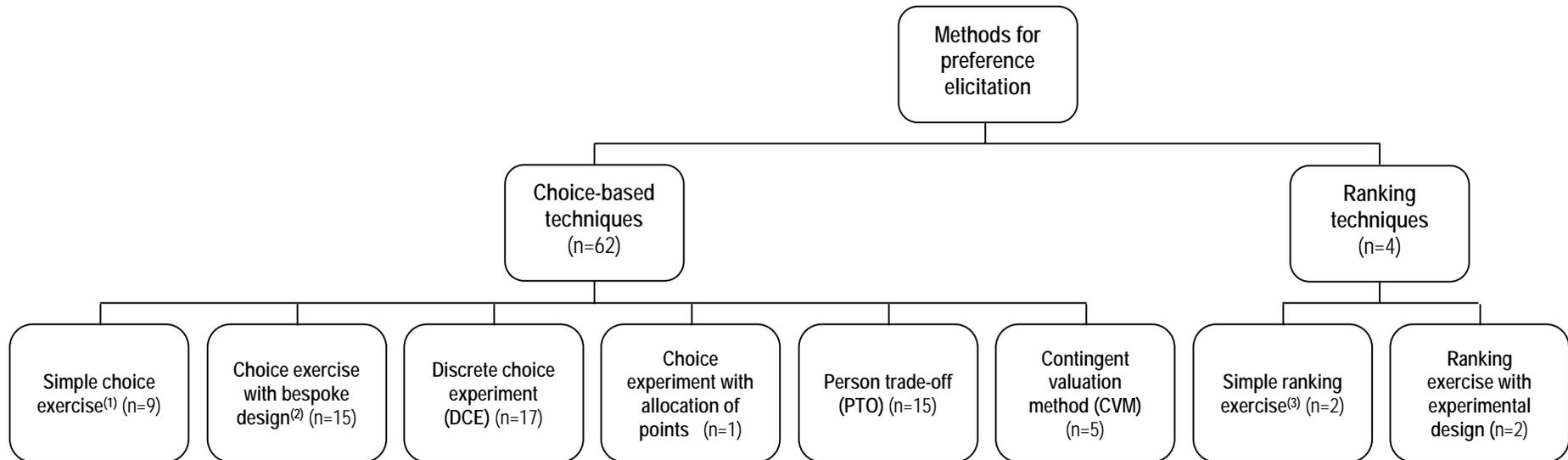


Figure 3 Categorisation of methods for preference elicitation



Note:

(1) Studies using *simple choice exercise* present respondents with scenarios that vary with respect to one attribute, and ask them to choose or allocate points between them. This category includes studies where the dependent variable is discrete choice or allocation of points.

(2) Studies using *choice exercise with bespoke design* typically involve multiple attributes and are designed to address specific hypotheses or theories without the use of a factorial experimental design.

(3) *Simple ranking exercise* ranks a list of attributes based on their relative importance.

(4) Frequency of use for each method is recorded in brackets. Whitty, et al. (2014b) used both DCE and BWS, and Pinto-Prades, et al. (2014) used both PTO and CVM. Hence, the numbers add up to 66 instead of 64.

Appendix 1 Complete search strategy

Database: Ovid MEDLINE, searched on 13 Aug, 2014

Database	Ovid MEDLINE(R) 1946 to Present with Daily Update	
Search No	Searches	Results
1	exp resource allocation/ or exp health care rationing/	15526
2	exp Health Priorities/	8892
3	(health care or healthcare).ti,ab.	295660
4	(resource\$ adj5 allocation).ti,ab.	7424
5	(priorit\$ adj5 set\$).ti,ab.	3289
6	3 and 4	2196
7	3 and 5	927
8	exp Technology Assessment, Biomedical/	9262
9	(health technolog\$ adj5 assessment\$).ti,ab.	1880
10	exp Drug Approval/ec, mt [Economics, Methods]	1055
11	(reimbursement adj5 decision\$).ti,ab.	430
12	(coverage adj5 decision\$).ti,ab.	525
13	(public insurance adj5 coverage\$).ti,ab.	57
14	cost effectiveness threshold\$.ti,ab.	253
15	1 or 2 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14	36876
16	preference\$.ti,ab.	88016
17	relative importance.ti,ab.	12932
18	(tradeoff\$ or trade off\$).ti,ab.	12549
19	league table\$.ti,ab.	228
20	(multi-criteria or multiple-criteria) .ti,ab.	832

21	exp Social Values/	18989
22	social value\$.ti,ab.	917
23	societal value\$.ti,ab.	263
24	distributional weight\$.ti,ab.	6
25	equity weight\$.ti,ab.	22
26	relative weight\$.ti,ab.	3102
27	(QALY\$ adj5 maximi\$).ti,ab.	41
28	(health adj5 maximi\$).ti,ab.	1087
29	value for money.ti,ab.	900
30	16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29	136739
31	15 and 30	2710
32	limit 31 to humans	2383
Outcome	2383 hits	

Database: EMBASE, searched on 13 Aug, 2014

Database	EMBASE	
Search No	Searches	Results
1	'health care'	1181803
2	'healthcare'	428490
3	#1 OR #2	1477019
4	priori* NEAR/5 set*	5104
5	resource* NEAR/5 allocation	22520
6	rationing	2793
7	#3 AND #4	2455
8	#3 AND #5	13860
9	#3 AND #6	2337
10	'health technology assessment'	4847
11	'health technology assessments'	428
12	'drug approval'	27653
13	reimbursement NEAR/5 decision*	957
14	coverage NEAR/5 decision*	791
15	'public insurance coverage'	43
16	'cost effectiveness threshold'	363
17	'cost effectiveness thresholds'	148
18	#7 OR #8 OR #9 OR #10 OR #11 OR #12 OR #13 OR #14 OR #15 OR #16 OR #17	51774
19	preference*	123200
20	'relative importance'	15837
21	tradeoff* OR 'trade off' OR 'trade offs'	15922
22	'league table' OR 'league tables'	299

23	'multi-criteria' OR 'multiple-criteria'	1322
24	'social value' OR 'social values'	1214
25	'societal value' OR 'societal values'	363
26	'distributional weight' OR 'distributional weights'	8
27	'equity weight' OR 'equity weights'	18
28	'relative weight' OR 'relative weights'	3606
29	qaly* NEAR/5 maximi*	65
30	health NEAR/5 maximi*	1814
31	'value for money'	1342
32	#19 OR #20 OR #21 OR #22 OR #23 OR #24 OR #25 OR #26 OR #27 OR #28 OR #29 OR #30 OR #31	161893
33	#18 AND #32	1647
34	#33 AND [humans]/lim	1219
Outcome	1219 hits	

Database: ECONLIT, searched on 13 Aug, 2014

Database	Econlit (via EBSCOhost)
Search syntax	((("health care" OR healthcare) AND (priorit* N5 set* OR resource* N5 allocation OR rationing)) OR (health technolog* N5 assessment* OR drug* N5 approval OR reimbursement N5 decision* OR coverage N5 decision* OR "public insurance coverage" OR "cost effectiveness threshold*")) AND (preference* OR "relative importance" OR tradeoff* OR "trade off*" OR "league table*" OR "multi-criteria" OR "multiple-criteria" OR "social value*" OR "societal value*" OR "distributional weight*" OR "equity weight*" OR "relative weight*" OR QALY* N5 maximi* OR health N5 maximi* OR "value for money")
Outcome	127 hits

Database: WEB OF SCIENCE (SCI-EXPANDED & SSCI), searched on 13 Aug, 2014

Database	WEB OF SCIENCE	
Search No	Searches	Results
1	TS=(healthcare OR "health care")	255941
2	TS=(priorit* NEAR/5 set*)	5256
3	TS=(resource* NEAR/5 allocation)	23089
4	TS=(rationing)	22299
5	#4 OR #3 OR #2	49932
6	#5 AND #1	3988
7	TS=("health technology assessment*")	1905
8	TS=("drug approval")	806
9	TS=(reimbursement NEAR/5 decision*)	486
10	TS=(coverage NEAR/5 decision*)	725
11	TS=("public insurance coverage")	37
12	TS=("cost effectiveness threshold*")	251
13	#12 OR #11 OR #10 OR #9 OR #8 OR #7 OR #6	7846
14	TS=(preference*)	199455
15	TS=("relative importance")	31097
16	TS=(tradeoff* OR "trade off*")	61920
17	TS=("league table*")	540
18	TS=("multi-criteria" OR "multiple-criteria")	8210
19	TS=("social value*" OR "societal value*")	3527
20	TS=("distributional weight*" OR "equity weight*" OR "relative weight*")	5101
21	TS=(QALY* NEAR/5 maximi*)	43

22	TS=(health NEAR/5 maximi*)	1082
23	TS=("value for money")	1255
24	#23 OR #22 OR #21 OR #20 OR #19 OR #18 OR #17 OR #16 OR #15 OR #14	304024
25	#24 AND #13	732
Outcome	732 hits	

Appendix 2 Summary of included studies

Study	Country	Sample	Perspective	Method	Attributes/ Stated Preferences	Distributional weights
Abasolo and Tsuchiya (2004)	Spain	973 respondents from Spain	Social non-specific / ex post	choice exercise with bespoke design	social class: low > high	
Abasolo and Tsuchiya (2013)	Spain	1013 respondents from Spain	Impartial decision maker / ex post	choice exercise with bespoke design	social class: low > high	
Baker et al. (2009)	UK	587 respondents representative of England population	Social non-specific / ex post	PTO (DCE is the same as Lancsar et al. (2011))	(1) age: overall the young are favored over the old; 20-40 years > 40-60 years > 0-20 years > 60-80 years (2) severity: not the most severe condition gets priority; instead, it is ranked among the lowest	Using the 'ratio of mean' method: age weights(average over severity) for 0-20, 20-40, 40-60, 60-80 years are 0.775, 1, 0.814, 0.527 severity weights (average over age) for 0->20%, 20%->40%, 40%->60%, 60%->80%, 80%->100% are 0.791, 1, 0.876, 0.848, 0.688

Bleichrodt et al. (2005)	The Netherlands	179 respondents from the Dutch population	Impartial decision maker / ex post	choice exercise with bespoke design; weights estimated using the social welfare function based method	expected lifetime QALYs: a new born with smaller lifetime QALYs is favored over a new born with larger lifetime QALYs	reported for different number of expected lifetime QALYs (see table 7 in Bleichrodt et al. (2005))
Bosworth et al. (2010)	US	1500 respondents each for two surveys, nationally representative	Social non-specific / ex post	DCE	(1) number of illnesses prevented or number of recoveries increased: its marginal utility from prevention policies is the same as the one from treatment policies (2) number of deaths avoided: its marginal utility from prevention policies is about twice as much as the one from treatment policies Both attributes have significant positive effects.	
Bryan et al. (2002)	UK	909 respondents from Hertfordshire, highly representative of local population	Social non-specific / ex post	DCE	(1) number of people (2) chance of success (3) LE after treatment (4) QOL after treatment All have significant positive effects.	

Charny et al. (1989)	UK	a random sample of 719 respondents from Cardiff	Social non-specific / ex post	simple choice exercise	(1) age: young children are less valuable than older children, but elderly people are less valued than young people (2) lifestyle/culpability: healthy > not healthy, inherited disease > self-induced disease (3) social class: no effect (significant minority prefers a director over a person without skills, a teacher over a lorry driver) (4) employment status: no effect (significant minority prefers employed over unemployed) (5) gender: no effect (significant minority prefer female over male) (6) marital status: married > single	
Corso et al. (2002)	US	1104 English-speaking US adults	Personal / ex post	CVM	prevention vs treatment: the former's WTP is larger (\$665 vs \$223)	
Cropper et al. (1994)	US	1000 households in Maryland (Maryland Poll), 564 households in the Washington D.C. metropolitan area, and a random sample of 1000 households	Social non-specific / ex post	PTO	(1) timing of health gain: today > future (2) age: the young are favoured over the old with a peak of age-related preference for patients aged around 28	For the median respondent, the weights for life saving in 100 years, in 25 years, and today are 1/45, 1/6, and 1; the weights for life saving at 20, 30, 40, and 60 years old are 8, 11, 7, and 1.

Diederich et al. (2011)	Germany	2031 respondents representative of the German adult population	Social non-specific / ex post	simple choice exercise (DCE is the same as Diederich et al. (2012))	age: little evidence that the German public accepts age as a criterion to prioritize health care services (strong evidence from DCE by contrast)	
Diederich et al. (2012)	Germany	2031 respondents representative of the adult population of Germany	Social non-specific / ex post	DCE	<p>Mean preferences:</p> <p>(1) health status: severe disease > light disease</p> <p>(2) quality of life: severely restricted > restricted > no restrictions</p> <p>(3) unhealthy life style: yes > no</p> <p>(4) age of patient: 43 > 25 > 68 > 87</p> <p>(5) family status: single with dependents > single without dependents >= couple with dependents > couple without dependents</p> <p>(6) occupational status: low > medium > high</p> <p>Relative importance:</p> <p>(1) health status (50%)</p> <p>(2) quality of life (24.7%)</p> <p>(3) age (12%)</p> <p>(4) family status (7.9%)</p> <p>(5) occupational status (4.6%)</p> <p>(6) unhealthy lifestyle (0.8%)</p>	

Dolan and Shaw (2003)	UK	a representative sample of 23 from York	Social non-specific / ex post	simple ranking exercise	no-treatment profile > health gain > time spent waiting for treatment > lifestyle choices > previous health profile > impact on others > claims based on compensation or reward	
Dolan and Tsuchiya (2005)	UK	100 respondents from Yorkshire and Humberside	Social non-specific / ex post	ranking exercise with experimental design	(1) age: 40 > 60 (2) severity: mixed results based on different severity measures: past health (QOL without condition): mixed - it had a significant effect in the context of imminence of death, but was not significant in the context of severity of health future LE without treatment: statistically insignificant future QOL without treatment: statistically insignificant age is the most important factor	
Dolan and Tsuchiya (2009)	UK	a random sample of 130 respondents from York	Social non-specific / ex post	choice exercise with bespoke design; weights estimated using the social welfare function based method	self-induced/unhealthy lifestyle: significant negative	respondents wanted to give people who have not cared for their own health about half as much weight as those who have cared for their health

Dolan and Tsuchiya (2011)	UK	130 respondents from York, broadly representative for the general population in UK	Social non-specific / ex post	choice exercise with bespoke design; weights estimated using the social welfare function based method	(1) SES: low > high (2) gender: no effect	SES: a given health gain in life expectancy to the lowest social class is weighted (by the median respondent) about seven to ten times as highly as an equivalent gain to the highest social class
Dolan and Tsuchiya (2012)	UK	600 respondents from England, broadly representative of UK population	Social non-specific / ex post	choice exercise with bespoke design; weights estimated using the social welfare function based method	age and health profile before treatment: value of health gain is larger for young people whose QOL is low	Reported in Table 1 of Dolan and Tsuchiya (2012), e.g., the general public value extra 1 year in full health for a 50-year-old who is otherwise about to die as about the same as extra 10 years in full health for a 70-year-old who is otherwise about to die.
Edlin et al. (2012)	UK	a sample of 559 respondents broadly representative of UK general population	Social non-specific / ex post	choice exercise with bespoke design	(1) lifetime health: the poorer are favoured (2) self-induced illness: disfavoured but its effect may be outweighed by the lifetime health effect	
Eisenberg et al. (2011)	US	2132 respondents from a nationally representative online panel	Social non-specific / ex post	PTO	age: the young are favored over the old	the weights for age 10 and 60 are larger than 10:1

Green (2009)	UK	261 respondents from Southampton City Council, broadly representative of local population	Social non-specific / ex post	choice exercise with bespoke design	<p>(1) Severity: In the severity of health question 60% indicated that a unit of health gain in a severely affected patient group was of greater social value to that same unit of health gain in a moderately affected patient group, all else equal.</p> <p>(2) SES: When described by level of disadvantage, 80% of respondents stated such a preference, which indicates that they attach a greater social value to a unit of health gain in a disadvantaged patient group, compared to a more advantaged group, all else equal.</p>	
Green and Gerard (2009)	UK	259 respondents from Southampton City Council	Impartial decision maker / ex ante	DCE	<p>(1) the average health improvement expected from the treatment: large > moderate > small > very small</p> <p>(2) cost-effectiveness of treatment – the value for money expected from the treatment: very good > fairly good > fairly poor > very poor</p> <p>(3) severity: yes > no</p> <p>(4) availability of other treatments: yes > no</p>	

Gyrd-Hansen (2004)	Denmark	a random sample of 3201 respondents from Denmark	Social non-specific / ex post	DCE	QOL(measured using EQ-5D health states) before and after treatment: patients in a more severe health state are favoured provided their expected benefits are large enough to bring them to the health level where their rival patients are without treatment - this equity concern was more significant on specific health dimensions such as pain/discomfort and anxiety/depression.
Gyrd-Hansen and Kristiansen (2008)	The Netherlands	a random sample of 2900 respondents from Denmark	Social non-specific / ex ante	DCE	Distribution of fixed gain: In the context of life-saving, it shows there are preferences for both spreading and concentration of health gains. Respondents also adopt thresholds when they value treatment offers (spreading is preferred if size of gain > 6 months life expectancy or probability of gains > 1/12). This result is consistent regardless of the perspectives.

Jelsma et al. (2002)	Zimbabwe	a random sample of 67 respondents from a high-density area of Harare	Social non-specific / ex post	PTO	age: the young are favored over the old, and 15 is the most highly valued age	The mean weights for 1, 15, 30, 45, and 75 years are (1) 1.76, 2.19, 1, 0.76, and 0.33 (when gain is stated as lifesaving) (2) 0.95, 1.67, 1, 0.97, and 0.43 (when gain is stated as sparing one year of illness)
Johannesson and Johansson (1996)	Sweden	a random sample of 1000 respondents from Sweden	Social non-specific / ex post	PTO	age: the young are favored over the old	The weights for life saving at 30, 50 and 70 years old are 1.0, 0.2, and 0.029.
Johannesson and Johansson (1997)	Sweden	a random sample of 1000 respondents from Sweden	Social non-specific / ex post	PTO	age: the young are favored over the old	Using parametric estimation, the weights for life saving at 30, 50 and 70 years old are 1, 0.13, and 0.025. Using nonparametric estimation, the weights for life saving at 30, 50 and 70 years old are 1.0, 0.2, and 0.029.
Johri et al. (2009)	US/Canada	a sample of 2009 respondents from an online panel broadly representative of US and Canada	Impartial decision maker / ex ante	simple choice exercise: allocation of points	age: people generally prefer allocating scarce resources to young patients over older ones but these preferences are significantly reduced when participants are encouraged to reflect carefully on a wide range of moral principles	

Lim et al. (2012)	South Korea	716 respondents from an internet panel broadly representative of Korea population	Not reported	DCE	<p>(1) QOL without treatment: low > high</p> <p>(2) life years remaining without treatment: short > long</p> <p>(3) survival gain after treatment: large > small</p> <p>(4) QOL gain after treatment: high > low</p> <p>(5) patient's household income level: low > high</p>	
Lancsar et al. (2011)	UK	a random sample of 587 from the adult population in England	Social non-specific / ex post	DCE; weights calculated using a compensating variation based method	<p>(1) age at onset: insignificant</p> <p>(2) age at death if untreated: significant (log-transformed, polynomial)</p> <p>(3) QOL if untreated: insignificant (QOL loss, log-transformed, polynomial), less severe is favoured</p> <p>(4) gain in life expectancy</p> <p>(5) gain in QOL with treatment</p> <p>The last two are combined as QALY gain, and participants preferred to treat patients who had larger QALY gains, but at a diminishing rate.</p> <p>end of life (implied): not favored</p>	<p>Weights for individual attribute levels (details in Table 4 of Lancsar et al. (2011)):</p> <p>Age of death without treatment: weights for 1,10,20,40,60,70, and 80 years old are 1.08,1.16,1.03,1,1.03, and 1.06;</p> <p>QOL loss: weights for 0.8, 0.9, 0.7, 0.4, and 0.1 QOL loss are 0.98, 0.96, 1, 1.04, and 1.03</p> <p>Weights for combination of multiple attributes (see Table 5 of Lancsar et al. (2011))</p>

Lewis and Charny (1989)	UK	a random sample of 721 respondents from Cardiff City	Social non-specific / ex post	simple choice exercise	age: overall the young are favoured over the old: 5>70, 35>60, 8>2	
Linley and Hughes (2013)	UK	4118 respondents recruited using an online panel, broadly representative of UK population	Social non-specific / ex post	choice exercise with bespoke design	<ul style="list-style-type: none"> (1) health gain: driving the choices, larger gain preferred (2) costs: driving the choices, higher costs preferred (3) severity of disease: favoured, regardless of health gain and costs (4) no alternatives: favoured, even if health gain is little (5) innovative treatment: favoured, if health gain is substantial (6) having dependents (informal carers): favoured, with health gain and costs fixed (7) children: not favoured (8) end-of-life premium: not favoured (9) disadvantaged population (e.g., low income): not favoured (10) rare disease: not favoured (11) cancer treatment premium: not favoured 	

Mortimer and Segal (2008)	Australia	a random sample of 271 from Australia	Social non-specific / <i>unclear</i>	DCE	(1) self-induced illness: no>yes (2) purpose of the intervention: prevention>cure (3) type of intervention: medical, lifestyle (4) number of lives saved per year: the more the better (the more effective one is favoured) (5) quality of evidence: strong>limited (6) cost of treatment: less costly is favoured (7) contributions from patients: less out-of-pocket contribution is favoured (8) age/life-stage: young children > young adult > working-age adult > older-age retiree	
Nord et al. (1993)	Norway	a random sample of 102 respondents from Norway	Impartial decision maker / ex ante	PTO	severity: more severe health state is valued higher	reported in Table 5 of Nord et al. (1993), e.g., saving one life is equivalent to returning 3 patients at the health state (112232) to full health
Nord et al. (1995)	Australia	119 respondents from Melbourne	Social non-specific / ex post	simple choice exercise: allocation of fixed budget	cost of treatment: not significant in determining priorities	

Nord et al. (1996)	Australia	176 respondents from Melbourne and four country towns	Impartial decision maker / ex post	PTO	(1) age: the young are favored over the old (2) size of health gain: the larger gain is favored but at a diminishing rate	Age weights are similar in two health gain scenarios: for 10, 20, 60 and 80 years, the median weights are 1.1, 1.0, 0.4 and 0.1. The discounted rates for health gain are calculated for different contexts and perspectives.
Norman et al. (2013)	Australia	552 respondents recruited from an online panel broadly representative of the Australia population	Social non-specific / ex post	DCE, weights calculated using the marginal rate of substitution	(1) gender: insignificant (2) smoking status: no>yes (3) income/SES: low income > high income (4) lifestyle: healthy > not healthy (5) carer status: carer > non carer (6) age of death/remaining LE if untreated: 45>60>75 (7) extra LE if treated (health gain): the larger the better but a diminishing effect	Weights for combination of attributes reported in Table 3 and 4 of Norman et al. (2013)
Olsen (2000)	Norway	716 respondents from the adult population in Norway	Citizen / ex post	choice exercise with bespoke design	Distribution of fixed gain: there is a threshold level of benefits to the larger group above which people prefer to distribute gains to as many people as possible but below which they prefer to concentrate gains.	

Olsen (2013)	Norway	a sample of 503 respondents broadly representative of Norway general population	Social non-specific / ex post	choice exercise with bespoke design	(1) age: the young are favoured (2) size of gain: large gain is favoured (3) end of life: little support for the "end-of-life" argument that a short life expectancy makes patients entitled to preferential treatment.	
Olsen and Donaldson (1998)	Norway	a representative sample of 143 from Troms county	Citizen / ex ante	CVM	type of health gains: life saving QALY gain > life extending QALY > QOL improving QALY (WTP/QALY: 2 NOK, 1.5 NOK, 0.2 NOK)	
Palanca-Tan (2013)	Philippines	a quota sample of 500 respondents from the five largest cities in Metro Manila	Social non-specific / ex post	DCE, weights estimated using the social welfare function based method	(1) age: 1-19 > below 1 > 20-59 > 60+ (2) number of patients: significant positive effect (3) life-years saved: significant positive effect	Based on model 1, the weights for life saving among age groups 'below 1', '1-19', '20-59', and '60+' are 4, 5, 3, and 1.
Pennington et al. (2013)	Denmark, France, Hungary, Netherlands, Norway, Poland, Spain, Sweden, UK	17657 respondents across nine European countries	Personal / ex post	CVM	(1) type of gains: value for life extending gains > value for QOL enhancing gains (2) end of life: modest premium indicated for end of life treatment	

Petrou et al. (2013)	UK	2500 respondents from an internet panel in UK	Impartial decision maker / ex post	PTO	age: the young are favored over the old, and 30 is the most highly valued age	Estimated weights vary according to different framings, age bases, and methods. In most cases, 30-year-olds have the highest mean weight.
Pinto-Prades et al. (2014)	Spain	813 respondents broadly representative of the Spain adult general population	CVM: Personal / ex post PTO: impartial decision maker / ex post	CVM and PTO	(1) end of life premium: yes (2) type of gain: at the end of life, quality of life improvement > life extension	The weights for end of life treatment and temporary health gain are around 1.5 to 1. The weights for QOL improvement at the end of life and life extension at the end of life are around 1.5 to 1.
Quintal (2009)	Portugal	70 respondents from two Portuguese municipalities	Impartial decision maker / ex post	PTO	geographical equality of health gain distribution: preferred but not at any cost	In the context of preventing children from diseases, the median respondent in both samples gives a weight of 0.8 to 1 for a health gain concentrated in region 1 and a health gain equally divided between the two regions.

Richardson et al. (2011)	Australia	a sample of 430 respondents broadly representative of Australian general population	Social non-specific / ex post	PTO, weights calculated as a function of TTO utility scores and severity measure (QOL loss)	(1) severity of pre-treatment condition: highly significant (2) size of health gain: positive but diminishing	See Table 7 of Richardson et al. (2011)
Richardson et al. (2012)	Australia	a sample of 532 respondents representative of Australia general population	Impartial decision maker / ex post	choice exercise with bespoke design	distribution of fixed gain: very strong preference for 'dispersion' of benefits rather than their 'concentration'	
Roberts et al. (1999)	UK	a random sample of 91 respondents, broadly representative of the population of a health district in south-east England	Social non-specific / ex post	DCE	(1) the chance of success of the intervention: 0.1%, 1%, 10%, 50% (2) the number of people receiving treatments: 1, 10, 100 (3) survival gains if treatment is successful: 1 year, 5 years (4) health state after treatment: good > poor (little support for health care programs that left patients in relatively poor health states even though this represented a prognostic improvement)	

					Results for the first three attributes were not reported.	
Ryynanen et al. (1996)	Finland	a random sample of 49 respondents recruited using a telephone directory	Social non-specific / ex post	DCE (called random paired scenarios in the paper)	(1) age: child > old (2) severity of disease: severe > mild (3) prognosis: poor > good (4) cost of treatment: expensive > inexpensive (5) self-induced: no > yes	
Schwappach (2002)	Germany	a sample of 127 respondents from an online survey in Germany	Impartial decision maker / ex post	PTO	improving patients' health > avoiding decline (cure > prevention)	The mean substitution rate between health improvements and avoided decline ranged between 0.47 and 0.64 dependent on the intervention. See more details in Table 3 of Schwappach (2002).
Schwappach and Strasmann (2006)	Germany	a random sample from an internet panel; 843 completed the first survey; among them 716 completed the second one	Impartial decision maker / ex ante	choice experiments with allocation of points	(1) age: children > employable age > teenager > seniors (2) combination of initial and post-treatment QOL: low-high > moderate-high > low-low > high-high (3) effect on life expectancy: plus 10 years >	

					plus 5 years > no effect > minus 5 years (4) frequency of the disease: common > rare (5) costs: below average > above average	
Shah et al. (2013)	UK	a quota sample of 50 respondents broadly representative of the UK general population	Social non-specific / ex post	choice exercise with bespoke design	end of life: weak evidence of support for giving priority to the patient with shorter remaining life expectancy At end of life, QOL improvement > life extension	
Shiroiwa et al. (2013)	Japan	a random sample of 2283 respondents from an online panel, broadly representative of Japanese general population	Personal / ex post	CVM	(1) severity: more severe health state is valued higher (2) end of life premium: no	
Singh et al. (2012)	UK	a sample of 1030 respondents representative of UK general population	Social non-specific / ex post	PTO	self-induced/unhealthy lifestyle: less weight was given to interventions related to self-induced illness than that is caused by health care or nature (genetic disorders)	The weights for genetic disorder, lifestyle disease, and sport injury are 1, 0.65, and 0.41.
Tsuchiya and Dolan (2007)	UK	271 members of the UK public	Social non-specific / ex post	choice exercise: bespoke design	social class: low > high (much stronger preference than the clinician sample)	

Tsuchiya et al. (2003)	UK	140 respondents from Yorks and Humberside (not representative on some variables)	Social non-specific / ex post	simple ranking exercise	age: overall the young are favored over the old	
Ubel (1999)	US	479 prospective jurors in Philadelphia	Q1,2,4,5: Social non-specific / ex post Q4,6: citizen / ex ante	simple choice exercise	severity of illness: more severe is favoured but affected by framing effects	
Ubel et al. (1996)	US	568 prospective jurors in Philadelphia	Social non-specific / ex post	simple choice exercise	distribution of fixed gain: a small gain to the many > a large gain to the few (strong preference for giving more a benefit)	
Ubel et al. (1998)	US	228 prospective jurors at the Philadelphia County courthouse	Social non-specific / ex post	choice exercise with bespoke design	(1) severity: significant preference for directing limited resources toward those with greater disabilities, regardless of whether those resources were targeted toward prevention or cure. (2) prevention vs cure: when the magnitude of benefit was held constant, the subjects slightly preferred prevention over cure.	

Ubel et al. (1999)	US	251 prospective jurors in Philadelphia (not representative but some variations in backgrounds)	Impartial decision maker / ex post	PTO	treatment potential: lifesaving to perfect health has the same value as lifesaving to pre-existing paraplegia but much larger value than lifesaving to onset paraplegia (treatment causes paraplegia) and lifesaving to avoidable paraplegia (treatment causes paraplegia which is avoidable)	lifesaving to perfect health vs lifesaving to pre-existing paraplegia vs lifesaving to onset paraplegia (treatment causes paraplegia) vs lifesaving to avoidable paraplegia (treatment causes paraplegia which is avoidable) = 1:1:50:5000
Ubel et al. (2000)	US	495 prospective jurors in Philadelphia	Social non-specific / ex post	simple choice exercise	distribution of fixed gain: preference for giving more people gain is affected by framing effects	
Ubel et al. (2001)	US	study1: 615 prospective jurors in Philadelphia; study 2: a sample of 68 respondents from the internet	Social non-specific / ex post	simple choice exercise	distribution of fixed gain: preference for giving more people gain is affected by framing effects and order effects	

Whitty et al. (2008)	Australia	a quasi-random sample of 161 respondents from Australia	Social non-specific / ex post	DCE	<p>(1) chance of success: significantly positive</p> <p>(2) average survival, for those in whom the new pharmaceutical is successful: significantly positive</p> <p>(3) average health-related QOL, for those in whom the new pharmaceutical is successful. If successfully treated, the recipient remains in this health state for the duration of their survival: No pain/discomfort > Moderate pain/discomfort > Extreme pain/discomfort</p> <p>(4) additional cost: significantly negative</p>	
Whitty et al. (2011)	Australia	a quasi-random sample of 161 respondents	Social non-specific / ex post	DCE	<p>(1) expected survival if untreated: short (3 months) > long (15 years)</p> <p>(2) expected health state if untreated: poor (extreme pain/discomfort) > moderate (moderate pain/discomfort and moderate anxiety/depression)</p> <p>(3) success rate: 60% < 90%</p> <p>(4) survival gain if treated: significantly positive</p> <p>(5) QOL if treated: significantly positive</p> <p>(6) cost: significantly negative</p> <p>(7) uncertainty (for decision makers only):</p>	

					high < low	
Whitty et al. (2014)	Australia	930 respondents from an online panel (broadly representative of Queensland and Australia population except on some variables)	DCE: Social non-specific / ex ante DCE (BWS): Social non-specific / ex ante	DCE, BWS	DCE: (1) benefit type: early diagnosis > prevention > survival improve > hospital waiting time reduction > QOL improvement > side effect reduction (2) good value for money: yes > no (3) alternative treatment available: no alternatives > upgrade of an existing intervention > an alternative but different intervention is already available (4) number of patients: 1000>=2000>500>10 (5) age: 35>10>60>85 (6) indigenous Queenslanders: yes > no (7) rural or remote area: yes > no BWS: reversal of preference ordering for some	

					attributes (benefit type and age)	
Winkelhage and Diederich (2007)	Germany	120 respondents from Bremen, broadly representative of local population	Social non-specific / ex post	ranking exercise with experimental design	<p>mean preferences:</p> <p>(1) age: 16 > 37 > 68</p> <p>(2) healthy lifestyle: yes > no</p> <p>(3) type of illness: acute > chronic</p> <p>(4) severity of illness before treatment: severe > light</p> <p>(5) improvement in health after treatment: large > middle > small</p> <p>(6) treatment costs: low > medium > high (not significant)</p> <p>relative importance:</p> <p>age: 20.9%</p> <p>severity of illness before treatment: 19.6%</p> <p>improvement in health after treatment: 19.5%</p> <p>healthy lifestyle: 15.3%</p> <p>treatment costs: 13.5%</p> <p>type of illness: 11.2%</p>	

Appendix 3 Summary of results on ‘other attributes’

Other attributes of beneficiaries

Four studies elicited preferences for beneficiaries who have dependents (Diederich, et al., 2012; Dolan and Shaw, 2003; Linley and Hughes, 2013; Norman, et al., 2013). Three suggest the general public tends to favour individuals with dependents over those without dependents (Diederich, et al., 2012; Linley and Hughes, 2013; Norman, et al., 2013). The other study ranked having dependents as one of the least important attributes (Dolan and Shaw, 2003). Two studies elicited preference for marital status. Charny, et al. (1989) suggested that married people were favoured over singles while Diederich, et al. (2012) found that singles were favoured over those who are married. Charny, et al. (1989) found both employment status and gender had no impact on resource allocation. Whitty, et al. (2014) reported indigenous people and those living in rural or remote areas were favoured.

Three studies elicited preference for QoL after treatment or the end health state (Bryan, et al., 2002; Roberts, et al., 1999; Whitty, et al., 2008). All suggest little support for health care programmes that may leave patients in relatively poor health states even though this represents a prognostic improvement. A similar attribute is treatment potential (i.e. the best possible end health state) and Ubel, et al. (1999) found that individuals with less treatment potential were not disfavoured even if their gain from treatment was very limited.

Two studies elicited preferences for the rarity of disease. Linley and Hughes (2013) reported that common diseases were favoured over rare diseases while Schwappach and Strasmann (2006) found that rarity of disease had no impact on resource allocation.

Two studies elicited preferences for lifetime QALYs (i.e., health from a specific time to death). Bleichrodt et al (2005) and Edlin et al (2012) both found people with fewer lifetime QALYs were favoured. Similarly, Dolan and Tsuchiya (2005a) elicited preferences for past QALYs (i.e., health experienced up to now) and found people with fewer past QALYs were favoured. Another study by Dolan and Tsuchiya (2005b) elicited preferences for past QoL (i.e., QoL of all the past years) and found people with less past QoL were favoured. All four papers interpreted their results using fair inlining arguments.

Other contextual attributes

Three studies elicited preferences for availability of alternative treatment and all found that a disease with no alternative treatment was given priority (Green and Gerard, 2009; Linley and Hughes, 2013; Whitty, et al., 2014). Linley and Hughes (2013) found this preference remained even when the treatment led to limited health gain. Other attributes include number of patients (3 studies: larger number is favoured) (Bryan, et al., 2002; Palanca-Tan, 2013; Whitty, et al., 2014), chance of success (3 studies: higher chance is favoured) (Bryan, et al., 2002; Whitty, et al., 2008; Whitty, et al., 2011), cost effectiveness (2 studies: more cost effective treatment is favoured) (Green and Gerard, 2009; Whitty, et al., 2014), and uncertainty in evidence (1 study: lower uncertainty in evidence is favoured) (Mortimer and Segal, 2008).

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