



Review article

Patient and general public preferences for health states: A call to reconsider current guidelines

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ABSTRACT

In economic evaluations of health care interventions, benefits are often expressed in terms of Quality-Adjusted Life-Years (QALYs). The QALY comprises length and quality of life into one measure which allows cross-disease comparability. The quality adjustment of the QALY is based on preferences for health states. An important normative choice is the question whose preferences for states of health we wish to capture. The answer to this question is directly related to the normative question regarding the appropriate maximand in health care decisions. Currently, preferences are commonly derived from the general public, rather than from actual patients. This choice, which can have large consequences on final outcomes of economic evaluations, has always been a topic of debate. This paper clarifies and furthers the discussion regarding the appropriate source of preferences for health state valuations, acknowledges the plurality of different perspectives, and argues that health economic guidelines could require analysis of benefit in terms of QALYs based on *both* patient and general public preferences.

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1. Introduction

Economic evaluations serve the purpose of supporting the process of allocating the health care budget in such a way that it optimally reflects a set of objectives agreed to by society or a relevant decision maker. In general, an economic evaluation identifies whether a new treatment increases welfare by considering whether its (incremental) benefits exceed its incremental costs. In the context of a fixed budget, the approach is often used to assert whether a new treatment gains more health per invested euro than the displaced activities due to the re-allocation of resources (Claxton et al., 2011).

In economic evaluations, effects of health care interventions are commonly measured and valued in terms of Quality Adjusted Life Years (QALYs). The QALY combines quality of life (morbidity) and survival (mortality) in a single metric. Preferences indicate the desirability of health states compared to being in full health and form the quality adjustment of the QALY. These preferences are typically elicited by asking respondents to imagine being in some impaired health state for a fixed period of time. Generally,

respondents are then asked to choose between this option and living shorter but in perfect health (Time Trade-Off) or between this option and a procedure restoring perfect health but with a risk of immediate death (Standard Gamble). Using such preferences, and anchoring the value of the state 'dead' at 0 and that of perfect health at 1, allows QALY scores for health states to be computed. These are subsequently used to determine health gains from treatment in an economic evaluation; the differences between the QALY score prior to treatment and that of the QALY score post treatment (or that between an intervention and a control group) determines the size of the gain (or loss) in quality and/or length of life. Preferences elicited from the general public (referred to here as 'general public preferences') are often considered to be most appropriate for use in health care decision making. However, there has always been considerable doubt that public preferences "tell the whole story" (Nord et al., 2005) and the issue remained a topic of debate throughout the past years (Dolan and Kahneman, 2008; Dolan, 2010; Dolan and Metcalfe, 2012; McTaggart-Cowan et al., 2011; Nord et al., 2009; Stiggelbout & De Vogel-Voogt, 2008).

International guidelines identify different sources of preference as appropriate for health care decision-making. In for instance the Netherlands and the United Kingdom, regulatory bodies prescribe preferences for health states used in the context of economic

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evaluations to be obtained from the general public. The Dutch guidelines justify this choice by stating that general public preferences are in line with the societal perspective, the insurance principle and that adaptation of patients to their condition might result in unfavorable cost-utility ratio's (Zorginstituut Nederland, 2015). In Sweden, however, patient preferences are preferred (The Dental and Pharmaceutical Benefits Agency (TLV), 2003). This choice can be of great importance: it has been shown that, on average, patients indicate a smaller impact of health impairments than expected by the general public (Krahn et al., 2003; Noel et al., 2015; Peeters and Stiggelbout, 2010; Zethraeus and Johannesson, 1999). This difference may be larger for worse health states (Burstrom et al., 2006), although there are studies reporting opposite effects (Lloyd et al., 2008; Pyne et al., 2009; Stolk and Busschbach, 2003), as well as studies that only report a difference when preferences are measured with a time trade-off task, rather than with a discrete choice task (Krabbe et al., 2011). In some instances using patient preferences may result in more favorable cost-utility ratios for interventions treating those patients than using public preferences (Oldridge et al., 2008), but this need not to be the case (Schackman et al., 2002). The effect of using patient or public preferences also depends on whether an intervention is purely quality of life improving or rather life prolonging, as we will highlight in this paper. With patient preferences we here mean preferences obtained in those who are actually in the health state under value and thus experience this health state.

The differences between general public and patient preferences, and the international differences in guidelines in this context, stress the need for an open and thorough discussion of the issue of source of valuation of health states to be used in economic evaluations. This paper aims to contribute to this discussion against the background of decision-making regarding the reimbursement of new health care interventions. We will start from the current practice in which the QALY, interpreted from either a welfarist or an extra-welfarist perspective (Brouwer et al., 2008), is the commonly used maximand (potentially somehow corrected for distributive concerns). In this paper, we focus on the question whose preferences should form the basis of this maximand. This discussion is to be seen against this decision making background, since the size of the identified benefit in economic evaluations may depend on normative choices regarding whose preferences for health states matter. We aim to further the discussion regarding health state valuations in the context of economic evaluations by introducing a way forward which includes the use of patient preferences alongside general public preferences. To that end, and given the dominance of the general public as source of health state valuation, we will revisit some main arguments for using general public preferences put forward in the debate. We will conclude that the current justification for the use of general public preferences for health states is, at best, incomplete.

2. Background

We first clarify some important concepts that are central to the discussion.

2.1. Patients and the general public

Patients are those individuals who are currently experiencing impaired health. 'Patient preferences' generally refer to patients valuing their own experienced health state with a direct utility assessment such as the Standard Gamble or the Time Trade Off method. Own health may either be expressed using some standardized descriptive method (e.g. EQ-5D descriptive system) or simply as 'own health'. Patients can also value other impaired

health states than their own (i.e. non-experienced health states), but this is not the focus of this paper. Following this definition, a group of individuals, who need not classify themselves as patients, but are experiencing a less than perfect state of health, are defined as a source of 'patient preferences' if they value their experienced state of own health.

General public preferences generally refer to the valuation by the general public of typically non-experienced health states, described in some useful way. Typically, this involves a more limited description of health than the open ended 'own health' that can be used in obtaining patient preferences (Stiggelbout & De Vogel-Voogt, 2008). A 'general public' sample will consist of both patients and non-patients, and thus cannot be considered a non-patient or 'perfectly healthy' sample (Dolan, 2000). To illustrate, in the Dutch general public sample used for the valuation of health states, described with the EQ-5D descriptive system, 21.1 percent of the population indicated to be in 'fair/poor' health (Lamers et al., 2006). Hence, a general public sample also includes current or former patients, as well as individuals who have experience with specific health states in their loved ones. Nonetheless, a representative sample of the general public is unlikely to contain large groups of individuals experiencing the health states commonly valued in health state valuations. Thus, the general public is asked to value a state of health they are mostly not experiencing at the moment of valuation, and which they typically never have experienced at all (although some might have).

2.2. Ex-ante and Ex-post information

In the context of health state valuations, ex-post preferences are obtained after/during an individual experiences illness, and ex-ante preferences are obtained before the experience of illness. According to proponents of the ex-post approach, public decisions, such as health care resource allocations, ought to be based on the "tastes of individuals in those states of nature that are realized" (Harris and Olewiler, 1979). In the current example, these are patients who are experiencing a specific health state under valuation. The ex-post position is contrasted with the ex-ante position, which suggests that societal decisions, like insurance decisions, ought to be based on the tastes of individuals about states of the world (here health states) that are not yet realized or currently experienced. In this case, that would be the general public. In 1890 the famous economist Alfred Marshall described the distinction in terms of desires and satisfactions in his influential 'Principle of Economics' as following:

"It cannot be too much insisted that to measure directly, or *per se*, either desires or the satisfaction which results from their fulfillment is impossible, if not inconceivable. If we could, we should have two accounts to make up, one of desires, and the other of realized satisfaction. And the two might differ considerably. For ... some of those desires ... are impulsive; many result from the force of habit; ... and many are based on expectations that are never fulfilled." (Italics in original. Marshall quoted in Joan Robinson (Robinson, 2006)).

In the context of health state valuations, however, the demarcation between ex-ante and ex-post valuations, or desires and satisfactions, is not entirely correct for two reasons. First, the general public also contains patients, and thus does not fully align with an ex ante position. Indeed, a purely ex-ante position would require a sample of only healthy persons or, at least, a sample with no one in the state under valuation. Second, as further explained below, depending on the valuation method used, preferences elicited from patients can also be considered to be 'ex-ante', since the elicitation

process itself also includes non-experienced health states (as it involves choices between at least two health states).

Furthermore, the question arises to which extent patient preferences fully align with an ex-post position. Either all observed health states in a study would have to be valued, or, when using a patient-valued preference-based instrument, the patient sample would need to consist of patients representing all observed health states in a study (theoretically as much as 3125 health states with the new EQ-5D-5L (Herdman et al., 2011)). Obtaining experience-based valuations for each of the 3125 health states would, of course, be rather problematic if not impossible. A Swedish valuation study of EQ-5D-3L health states used experience-based (i.e. patient) preferences for 148 out of 243 potential health states as input for a regression model to estimate the values of the unobserved health states (Burström et al., 2014). By comparison, the Dutch EQ-5D-5L valuation study used preferences of the general public for 86 health states to estimate the values of the full set of 3125 health states (Versteegh et al., 2016).

2.3. Preferences, decision utility and experienced utility

Preference is a general term that is frequently used indistinctively to describe all sorts of valuations and orderings of alternatives. It is possible and relevant, however, to distinguish between different types of preferences. The preference one holds for states of the world before they actually occur can be labeled as decision utility which can be meaningfully differentiated from the experienced utility once one is in that state of the world (Kahneman and Sugden, 2005). Preferences for health states, whether for own health in patients or for hypothetical health states in the general public, are typically elicited in choice based valuation studies. The choices individuals make in such stated choice experiments indicate the order and strength of preferences for certain aspects of health. According to Kahneman & Sugden (Kahneman and Sugden, 2005), any preference elicitation, whether from patients or the general public, is (partly) ex-ante information as the elicitation process itself involves choices between health states of which at least one state is hypothetical, given that one can only be in one state of the world (i.e. health state) at a time. In a Time Trade-Off exercise, for example, respondents are asked to choose between living in an impaired health state for a period of time t or living in full health for a period of time x where $x < t$. From the perspective of the (majority of the) general public, the impaired health state will be hypothetical while full health is the experienced state, but from the perspective of the patient full health will be hypothetical while the impaired state (when valuing own health) is experienced. Therefore, from both perspectives the stated choice design produces information (partly) based on ex ante preferences (decision utility) and partly based on ex post preferences (experienced utility). While common health state valuations may mix decision and experienced utility, the obtained valuations do differ considerably when elicited in the general public rather than in patients (Peeters and Stiggelbout, 2010). This difference may have important consequences for calculations of the size of health gains, which, in economic evaluations, is based on the valuation of health. In this paper the terms 'preference' and 'utility' are used to refer to the value individuals place on a state of health.

2.4. Valuation of health and effect measurement

The valuation of health refers to the process of assigning a preference or a 'utility number' to a certain state of health (Brouwer et al., 2008). The measurement of health benefits due to a treatment refers to pre- and post-therapy differences (or those between a treatment and a control group) in the health states of patients,

which can be subsequently quantified by attaching utility numbers to both health states and calculating the difference, accounting for life duration, in terms of QALYs. When general public preferences are used, effect measurement is a process distinct from the valuation of health, often referred to as an 'indirect method'. Then, commonly, effect is measured by letting patients indicate their health state using a descriptive system (e.g. a questionnaire such as EQ-5D). Each health state in the descriptive system has a preference score attached to it, separately elicited from the general public. The magnitude of effect of a treatment is then defined by comparing the health states (accounting for length of life) of patients pre-therapy (or control group) to those in post-therapy patients (or intervention group), using the obtained preferences to calculate average QALY changes.

When using the patient perspective, preferences are generally (but not necessarily) elicited for own health. In that case effect-measurement and the valuation of health can be performed in one and the same process. This may therefore be referred to as a 'direct method'. Nonetheless, patient preferences could equally well be part of an 'indirect method'. This is the case when the health states of a questionnaire, such as EQ-5D, were previously valued by those who experiencing the valued health states (i.e. patients), after which these valuations were used to obtain a value set. This has been referred to as an 'experience-based value set', as opposed to a general public value set (Burström et al., 2014). Once this value set exists, the indirect method can be applied using patient preferences as well. (It needs noting that when patient health state preferences elicited with the direct method are compared to general public preferences for health states elicited with an indirect method, potential differences may relate both to the source of preferences as well as to methodological differences.)

It is important to note that measuring changes in health states using a descriptive system (like EQ-5D or HUI-III) is commonly performed in the affected patients. As we will discuss later, adaptation to one's health state may not only influence the value attached to health states, but may also influence the way in which descriptive systems are interpreted and completed. At least, it is important to distinguish between the valuation phase and the measurement phase, whenever relevant.

Above, the main concepts that are relevant to the debate about "who should value health" were clarified. Now we proceed with the debate itself, which we categorized in three key subjects, which align with the main arguments in favor of using general public preferences, as provided in the Dutch guideline for economic evaluations: the societal perspective, adaptation (with some related arguments) and the insurance principle.

3. Revisiting the arguments

Without claiming to be exhaustive, we discuss some of the main arguments put forward in favor of using public preferences below. We will argue that none of these arguments, alone or jointly, provides a convincing argument for solely using either general public preferences or patient preferences.

3.1. The societal perspective

The influential Panel of Cost-Effectiveness in Health and Medicine (Gold et al., 1996) (hereafter 'The Panel'), stated that "for purposes of resource allocation, the relevant preferences are those of the general public" (p.111). The following section draws heavily on this handbook, since it extensively and influentially addressed this important topic. The Panel links the claim that general public preferences are the relevant ones to the argument that cost effectiveness analyses ought to adopt a societal perspective. Public

preferences are said to be “a logical extension of the societal perspective” (p.99). The societal perspective normally relates to the scope of costs and benefits considered in economic evaluations, like Gold et al. (1996) define:

“When a CEA is conducted from the societal perspective, the analyst considers everyone affected by the intervention and counts all significant health outcomes and costs that flow from it, regardless of who experiences the outcomes or costs” (Gold et al., 1996) (p 6).

Following the societal perspective, costs and effects beyond those directly related to the patients are considered of importance, such as productivity costs (Koopmanschap et al., 1995) and measuring health effects in and costs for informal caregivers (Hoefman et al., 2013). It is not entirely clear, however, why adopting this broad perspective which incorporates everyone affected by the intervention would suggest that the general public is the most appropriate source of preferences. At most, it suggests that preferences of, and effects in, others than patients matter too.

According to the panel (Gold et al., 1996), the quality adjustment part of the QALY has to be based on decision utilities (rather than experienced utilities), and community samples of the general public are the appropriate source of preferences to make comparisons across interventions and populations (p.99).

“One way to see the desirability of the societal perspective ... is to imagine for a moment that we are looking at the world before we are born, or at least before we encounter any serious health problems, and to ask what kind of world we would like it to be. In that “ex-ante” position we would not yet know what sort of health problems we were destined to develop ... we might reasonably prefer a system in which decisions about health interventions reflected the seriousness of the problem and the ability of the intervention to do something about it, without reference to the specific individuals with the problem or to particular budgets or special interests”. (p.6 & 7).

Since economic evaluations aim to meet a set of societally agreed objectives, it is important that these objectives do not favor the preferences of special interest groups. Therefore, preferences that drive outcomes of economic evaluations ought to be elicited behind a veil of ignorance. One can doubt whether the general public reflects Rawls' ‘original position’ where “no one knows his place in society, his class position or social status ... and the like” (Rawls, 1999). In that context, however, one could state that a specific part of the general public, i.e. healthy individuals, would be the relevant source of information for health state valuations (which is the topic in this paper, rather than societal decision making *per se*), which does not align with the current sampling strategy which includes individuals with health problems. However, it has empirically been shown that the low number of patients in a general public sample does not affect the preference-based value set (Jonker et al., submitted).

Whether one, even behind the veil of ignorance, would not want to know how health states are actually experienced, seems unclear. Thus, if the societal perspective refers back to the Rawlsian original position, it seems that the assertion that a sample from the general public would be the ‘logical’ source of preferences is perhaps a bit too simple, since it may equally well be that behind the veil of ignorance, we might reasonably prefer that the seriousness of the problem and the ability of an intervention to do something about it is based on values of those who have firsthand experience with these matters, i.e. patients. Then, the question remains whether patients' valuation of a health state, captured with preference

elicitation methods, are not biased.

3.2. Adaptation

Adaptation can be defined as ‘to adjust oneself to new or changed circumstances’ (Webster's New World Dictionary, cited in Menzel et al. (Menzel et al., 2002)). Menzel et al. (2002) nicely summarize the issue of adaptation in the context of choosing between patient and general public values to inform resource allocation:

“No matter how sad the circumstances are in which disabled and chronically ill people adapt, we would seem to be ignoring their laudable adaptation were we merely to proceed as if they had not adapted. Yet we may also doubt that we are fully respectful of disabled persons if we proceed simply on the basis of their own values, forgetting the circumstances which caused them to adapt.” (Menzel et al., 2002).

3.3. Adaptation: hypothetical health states

The use of preferences obtained from the general public “might be reflecting the objective fact that the range of capabilities for people having certain conditions and disabilities is lessened compared to the normal range” (Gold et al., 1996) (p.99). The general public is generally healthy and can judge the loss of capabilities from the viewpoint of someone who is in full health. This may result in a ‘better’, or at least uniform, representation of the ‘distance’ between being in full health and having the health impairment. For patients the reference point they are reasoning from may have shifted and full health may be too hypothetical (Brazier et al., 2007), which may be considered problematic when wishing to come to universally applicable health state valuations. On the other hand, for the general public impaired health may be too hypothetical, causing the general public to overestimate the impact of health impairments yielding rather low, and perhaps relatively often negative, that is ‘worse than dead’, preferences which indeed are not observed in the Swedish EQ-5D value set based on respondents who value their own health (Burström et al., 2014).

The differences between patient and public preferences for health states seem to indicate that the general public may not adequately forecast experienced utility of being in a health state, and are, in that sense, ‘wrong’. Fitzpatrick writes:

“individuals appear to make value judgements about the desirability or undesirability of hypothetical health states by focusing on the transition from their own current state to the imagined hypothetical states” (Fitzpatrick, quoted in Sharma et al. (2004)).

The focus on transitions may lead to beliefs about the impact of disease by the general public that is not reflected in experiences of patients (Dolan and Kahneman, 2008). Indeed, when uninformed, there may be a general misunderstanding of what it is like to live with a disability (Ubel et al., 2003), at least when this disability is presented to the general public in the form of a health state in a valuation task. This does not mean that patient preferences are preferable to general public values, only because the general public might not adequately forecast adaptation one might experience in a health state. There may be several complex mechanisms involved that cause differences between health state preferences derived from patients and the general public, and not all of these mechanisms reflect information that decision-makers would wish to

include in economic evaluations. In their article, [Menzel et al. \(2002\)](#) describe eight elements of adaptation that argue either for or against using preferences shaped by adaptation. ‘Cognitive denial’, ‘suppressed recognition of full health’ and ‘lowered expectations’ are three forms of adaptation which might not be adequately forecast by the general public, but these are arguably perception biases in patients rather than an ‘adjustment of oneself’ following from a complete understanding of changed circumstances. According to the authors, recognizing that these three elements can cause researchers to observe ‘adaption’ in patients argues against using preferences shaped by adaptation. On the other hand the authors note that, ‘skill enhancement’, ‘activity adjustment’, ‘substantive goal adjustment’, ‘altered conceptions of health’ could be considered ‘laudable adaptation’, and reflect actual ‘adjustment of oneself’ rather than perception biases and may argue for including adaptation effects in decision-making. Indeed, Menzel and colleagues conclude that adaptation in itself is not a reason to disregard patient preferences, perhaps only when “comprised primarily of cognitive denial or suppressed recognition of full health (...)” They proceed to conclude the following:

“If, for example, the disabled and chronically ill themselves, after lengthy discussion of this issue, were to doubt that most values influenced by adaptation should be used in setting priorities among health services, the case for using actual patient values might be weakened considerably.”

Below we clarify how adaptation might impact the result of cost-utility studies, keeping in mind that its impact on the outcome of a cost-utility study is a consequentialist argument. Nevertheless, the ‘emotional argument’ that general public preferences are required because adaptation would result in an underestimation of effect has a strong appeal and therefore deserves a thorough discussion.

3.4. Adaptation: underestimation of effect

In the words of Cohen, when explaining the critique of Amartya Sen on the informational base of utility:

“The fact that a person has learned to live with adversity, and to smile courageously in the face of it, should not nullify his claim to compensation” ([Cohen, 1993](#)).

Indeed, one of the perhaps most influential reasons for favoring the general public (or rather healthy respondents), as source for health state valuations is that the “gains possible from the intervention become larger when the perspective is that of the general population” ([Gold et al., 1996](#)) (p.102). It is actually uncertain whether there is a ‘larger gain’ when using general public values. First, the direction of the effect of adaptation has contradicting empirical findings, as referenced in the introduction. Second, even when patients are assumed to produce higher values, this does not necessarily result in ‘larger gains’. Adaptation may affect the size of the effect of successful treatments in several ways, as explained below, with important implications for resource allocation in health care. Regarding curative interventions which improve quality of life without prolonging length of life, the ‘gain’ from an intervention may be smaller with patient values: fully curing a patient who identifies herself as rather healthy results (e.g. 0.8) in a smaller gain (i.e. 0.2) than when using lower general public values for the same health state (e.g. 0.6 and 0.4 respectively).

However, for life extending interventions this may be different. The ‘gain’ from an intervention may then be *higher* when valued by

patients. A higher quality of life weight in life years gained results in higher health gain in. This could, for example, be relevant in cancer patients ([Arnold et al., 2009](#)). Using patient preferences, any additional life time receives ‘more weight’ than with general public preferences and hence the QALY gain is larger. Therefore, **general public preferences do not always protect patients against the consequences of their adaptation.**

Moreover, even in case of interventions that increase quality but not length of life, using patient preferences may increase health gains. This depends on the nature of the adaptation process. Imagine a very poor health state, valued rather low by general public and patients, 0.3 and 0.4 respectively. Treatment can bring these patients to a better, but still impaired, health state, to which patients may adapt more easily, for example, going from a hospital bed to a wheelchair (see [Table 1](#)). The new health state (the wheelchair) may allow a patient to have a relatively large ‘adaptation opportunity’ compared to the old health state. In other words, the magnitude of adaptation can be asymmetrical in size pre and post therapy, due to the nature of the condition pre and post therapy. At present, it appears largely unknown when and with which magnitude adaptation occurs in impaired health. This (theoretical) example shows that using patient preferences which include adaptation does not always result in less favorable incremental cost-utility ratios in curative rather than life prolonging settings and indicates that the ‘adaptation’ argument for using general public preferences is perhaps less straightforward than sometimes assumed.

In sum, using general public rather than patient preferences can be advantageous as well as disadvantageous for patients, depending on the specific circumstances. Identifying these circumstances is of crucial importance for a better understanding of the real effects of (ignoring) adaptation in cost-utility analyses in public decision-making.

3.5. Adaptation: an aside on descriptive systems

In current practice the outcome of interventions is assessed using a preference-based descriptive system (i.e. a questionnaire with an algorithm attached to it). Although issues with adaptation often lead health economists to favor public preferences ([Verkerk et al., 2001](#)), the current two-stage assessment system with a questionnaire and a separate algorithm to compute preferences of the general public does not necessarily rule out adaptation fully. Adaptation may also be ‘picked up’ by the descriptive system of a questionnaire ([Brazier et al., 2007](#)). The elderly, for example, might report that they do not ‘have problems’ with their mobility or usual activities as they consider their somewhat declined mobility or shifted usual activities to be normal *for their age and peer group*. The same elderly individual will probably not state that s/he is able to walk 5 km without resting, suggesting that the phrasing of the question itself might also induce sensitivity to adaptation despite the presence of a value set elicited from the general public. Thus, **next to the valuation phase, descriptive systems may be sensitive to adaptation.** As a result, using public preferences to value health states of a descriptive system may not be a complete solution to the issue of adaptation.

Table 1
Asymmetrical adaptation.

	General public preferences	Patient preferences
Pre treatment	0,3	0,4
Post treatment	0,6	0,8

3.6. The insurance principle

Some have argued that the “insurance principle” (Hadorn, 1991), is an argument for using public preferences to value generic health states:

“But whose preferences should be used to determine the *value of a treatment* for purposes of societal allocation rules? Patients who rely on others to pay their medical bills ... cannot expect that these others will pay for everything they (the patients) might wish to receive. Permitting patients unlimited access to care based on post-illness preferences would too often result in the provision of marginally beneficial care. The lack of any associated marginal financial cost to the patient often makes any potentially beneficial treatment desirable or ‘worth trying’. For this reason, the importance and priority of treatments should be based on the average pre-illness preferences of the entire beneficiary population ...” (Hadorn, 1991).

The insurance principle, therefore, argues that having insurance is likely to alter sensitivity to the cost of treatments and that, in absence of opportunity costs to the individual patient (given insurance), experiencing illness may result in inelastic demand for treatments. Consequently, patients may desire marginally beneficial care despite high costs of care, as they are insured against those costs and share the burden of additional costs of treatments with the entire insured population. When patient preferences would be used to determine the *content of insurance packages*, the price attribute of treatments would possibly be disregarded by the patients. The insurance principle points out that, as a consequence, *there would be no sensible constraint on the provision of marginally beneficial care, and the insurance premium, paid both the patient and the entire insured population, would soar.*

This, of course, may be completely correct, but is *not very relevant to the question at hand here, as it does not apply to the current use of preferences in cost-utility analysis.* In cost-utility assessments, preferences are elicited for health states. These preferences are subsequently used to indicate the effect of treatment, separately from both the costs and the monetary valuation of health changes. In a later stage, mostly without direct patient involvement, the marginal societal costs of treatments are divided by marginal preference based effects, relative to some comparator, and compared to some relevant societal willingness to pay threshold for health gains. Given how preferences are commonly used in economic evaluations, the insensitivity to costs of treatments induced by insurance and in some cases the decreased marginal utility of money (e.g. in life threatening situations) is irrelevant for the question whose values count in valuing health states. Patients would only be called upon to value health states and to indicate in which health state they are in different stages of the illness and treatment. This information is then used to calculate the effect a treatment, but the desirability of *a treatment itself* is not, in any direct sense, asked.

Another interpretation of the insurance principle, “found routinely in other areas of insurance” (Hadorn, 1991), may be considered more compelling. An individual chooses an insurance package, in an ex ante situation, under uncertainty about a future state of the world, such as the likelihood of an adverse event like an earthquake that may damage property, or, when applied to health insurance, the likelihood of needing treatment. Since the individual chooses a specific health insurance package ex ante (since in many contexts burning houses are hard to insure), the logical thing would be to also use ex ante preferences in deciding on the package. Moreover, since in many Western countries all individuals are (obliged to be) insured, it is also logical to use the preferences

existing in a representative sample of the general public. However, two points can be made here indicating that patient preferences can still be relevant. First of all, while individuals and collectives often have to decide ex ante on insurance packages, this does not mean that they would always opt for the use of own ex ante preferences regarding health states. It is well conceivable that informed citizens would opt to use ex post preferences that arguably better reflect the actual impact of illness on well-being, if this were possible. Second, in a collectively financed health care system, the ex ante position holds for most insureds and payers, but not for all. One may well claim that the system is constructed (using solidarity principles) in such a way as to maximally contribute to the avoidance and relief of *actual* losses of well-being, rather than expected losses and such losses can be observed in those individuals at which the health care system is primarily targeted, i.e. patients.

If the insurance principle is taken to mean that those who pay for health care (through insurance premiums) should be able to participate in policy decision concerning health care, it has to be acknowledged that such involvement may take many other forms than health state valuations. Examples include citizen panels, democratic deliberation on the content of the basic benefit package or studies into the preference of the general public for distributive concerns. This does not preclude the use of ex post patient preferences for health states. Hence, while providing important arguments in the debate regarding whose values (should) count, the insurance principle, regardless of the specific interpretation, does not seem to provide a decisive argument in favor of ex ante, public preferences for the valuation of health states.

3.7. Taxonomy of arguments

The arguments discussed above, for and against the use of one specific source of health state values, can be grouped into normative/theoretical and empirical issues. Mainly normative arguments are the ones related to the societal perspective and the veil of ignorance, as well as the different arguments based on the insurance principle. None of those were found to be convincing as a rationale for strictly adhering to general public preferences. *Empirical arguments mainly relate to adaptation and the several issues arising from that phenomenon.* It is important to gain more knowledge of adaptation and its influence on health state valuations. Again here, these arguments in favor of either general public or patient preferences, do not form a decisive argument for either source. A taxonomy is provided in Table 2 below, stating the main line of reasoning, the nature of the argument and the main counter argument (be it simplified).

4. Way forward

Although the properties of both the public and the patient preference perspectives have been extensively discussed in the literature, there does not seem to be any conclusive or theoretically sound justification for disregarding either perspective, as has been argued before (Drummond et al., 2009; Menzel et al., 2002; Menzel, 2014). One suggestion is that *“members of the general public could be informed about the views of those who have experienced the health states that are the subject of valuation”* (Drummond et al., 2009), which aligns with a previous suggestion to provide the general public with “more information on the size and the nature of the adaptation experienced by patients over time” (Brazier et al., 2007). At least three outcomes are imaginable from such a procedure. First, the general public, once informed, might be convinced of the ex-post perspective of the patient and hence general public valuations will be equal to patient valuations. Second, the general public could completely ignore information on experienced utility,

Table 2
Taxonomy of argument for and against societal and patient preferences for use in health valuation.

Argument	Key line of reasoning	Nature of the argument	Main counter argument
Societal perspective	Public preferences are a logical extension of the societal perspective	Theoretical (GP)	Societal perspective can also include patient preferences
Veil of ignorance	Societal choices should be 'impartial'; not favouring particular groups or interests. Public preferences do not favor particular groups or interests.	Normative (GP)	Knowing real impact remains important, also behind a veil of ignorance.
Adaptation (1)	The general public is generally healthy and can judge the loss of capabilities from the viewpoint of someone who is in full health. This may result in a uniform representation of the 'distance' between being in full health and the valued health state.	Normative/Empirical (GP)	Uniformity is not the sole relevant criterion nor is it synonymous with correct.
Adaptation (2)	For the general public impaired health may be too hypothetical, causing the general public to give too low preference scores, overestimating the impact of poor health on wellbeing.	Normative/Empirical (P)	All choices contain a hypothetical comparator.
Adaptation (3)	General public preferences protect patients from the negative distributional consequences of their own adaptation.	Normative/Empirical (GP)	Use of GP valuations can also be against patients' interests.
Insurance principle (1)	Absence of financial consequences of health care use for patients could lead to a preference to use any potentially beneficial treatment.	Normative/Empirical (GP)	Relation with current (use of) health state valuations unclear.
Insurance principle (2)	Individuals choose a specific health insurance package ex ante, which aligns best with ex ante preferences.	Normative (GP)	Choosing on the basis of ex post information may still be preferred.
Insurance principle (3)	Those who pay for health care (through insurance premiums) should be able to participate in policy decision concerning health care.	Normative (GP)	Such participation may take many forms and does not preclude use of ex-post preferences.

GP: argument for general public preferences

P: argument for patient preferences

so that their ex-ante valuations remain unaltered. Third, the informed ex-ante position could be partially modified – taking some intermediate value for all health states, or affecting only some health state valuations, but not all. In all three cases, the final outcome is still one value per health state, which facilitates the decision making process. However, whether this value, at best a mix of both 'perspectives', is considered the most appropriate way of dealing with the issue at hand may be questioned. One could view the outcome to result in a loss of information, since exactly the *discrepancy* between QALYs derived from general public and patients is relevant information to decision-makers, as long as this discrepancy is considered to be caused by a valid difference of opinion regarding quality of life in a state of health.

Another suggestion is to **separate the societal value of health gains and the value of health states** (Ubel et al., 2000). In such an approach, societal preferences for distributive concerns regarding health gains are estimated with the Person Trade-Off technique in the general public, and the actual health state values are elicited in patients using some other elicitation technique (i.e. Time Trade-Off). While this suggestion is probably the most elaborate attempt to distinctively include both general public and patient preferences, it is not entirely congruent with the issue at stake here. The Person Trade-Off may pick up distributional concerns for health and health care in the general public, which goes beyond the mere utility value of being in a certain health state as experienced or expected by an individual. Indeed, the original authors explicitly acknowledge that an underlying assumption in their approach is that patients "have the best information concerning disutility of different health states". Hence, following this approach does not directly address or solve the issue discussed in this paper.

4.1. A solution involves both perspectives

The most elegant solution may be to include both the ex-ante and ex-post position. Historically, the disadvantages of patient preferences and advantages of general public preferences appear to have been emphasized, resulting in (or perhaps even following from) a large practical consensus to use generic measures of quality of life which have general public preferences attached to it. However, it seems difficult to claim that patient preferences, which are arguably better informed concerning the actually experienced loss of health related quality of life in a health condition, do not carry important information for societal decision making. Indeed, public preferences provide a reliable and systematic standardization for the computation of QALYs, and may represent the ex-ante assessment of the likelihood (or difficulty) of an adaptation process, but do not adequately forecast the experience of physical and mental impairment due to shifting preferences and, potentially, a lack of experience with impairments.

Reflecting on the different arguments put forward in the literature, it seems that both public and patient preferences hold information on health related quality of life that is potentially relevant for societal decision making. Consequently, **neither public nor patient preferences can be the 'royal road' to the evaluation of health care interventions in themselves.** One may wonder whether the two positions are necessarily mutually exclusive. To date, the debate seems to have centered around the question which of the two perspectives would be most appropriate. It seems unclear, however, why such a dichotomy would be necessary or, in fact, useful. This helps decision makers in recognizing the consequences of applying either perspective. To quote Sen:

“Euclid is supposed to have told Ptolemy: ‘there is no ‘royal road’ to geometry’. It is not clear that there is any royal road to evaluation of economic or social policies either. A variety of considerations that call for attention are involved, and evaluations have to be done with sensitivity to these concerns.” (Sen, 1999) (p.85)

For the evaluation of health care interventions it may be important to acknowledge that while the prospect of some impaired health states may seem extremely undesirable to the general public, it may be experienced as less undesirable once experienced by actual patients. These two considerations, both before and after health impairments, seem relevant in their own right in decision making.

Indeed, it seems difficult to argue why a societal decision maker, faced with a choice between two programs which are fully identical in all respects but the ex post patient health state valuation would not, and even should not, use this information to allocate the money there where less adaptation is possible. This can be highlighted with two examples. First, when decision makers have to prioritize one of two programs, which are equal in all aspects but the burden of illness from the perspective of the general public, which is greater in program A, would they not logically prioritize program A over program B, as in this program the burden of illness from the perspective of the general public is higher (i.e. a lower quality of life)?

Second, when decision makers have to prioritize one of two programs, which are equal in all aspects, except that in program B patients are more prone to adaptation than in program A, would they not logically prioritize program A over program B?

The fact that patients may adapt to certain health impairments (to different extents) need not be seen as disqualifying patient preferences, but rather stress the importance of including them in societal decision making. Still, some have argued that adaptation (and the use of preferences affected by adaptation) results in undervaluation of the benefit of curative treatments. It seems that this ‘protecting the patient against own adaptation’ argument has received most attention so far, even though it is only a part of the full story as explained above. Often disregarded, not empirically confirmed, but nonetheless of importance, is that the advantage of using public preferences –the potential insensitivity to adaptation– may currently not even be achieved as the descriptive systems to which societal preferences are applied could be sensitive to adaptation. People may for instance have different appreciations of terms like ‘usual activities’ and mobility, when in a poor health state, influencing health state descriptions.

To make sure the differences between sources of preferences are not due to differences in methodology, it is crucial that the methodology of measuring patient preferences aligns with the methodology for eliciting general public preferences: valuation exercises for a generic description of a state of health. To overcome feasibility issues, it is acceptable to limit these exercises to the most commonly observed health states of, for example, EQ-5D, or to estimate the value of the unobserved health states with regression techniques, as has been done before (Burström et al., 2014).

4.2. A proposal to rethink current guidelines

One way forward for the evaluation of health care interventions

is to acknowledge the plurality of relevant perspectives rather than advocating only one ‘royal road’. To that end, health economic guidelines could require analysis of benefit in terms of QALYs using both patient and general public preferences, effectively requesting that the reference-case analyses consists of two ICERs. This is similar to earlier pleas for presenting ICERs from two perspectives (i.e. the health care and societal perspective). Indeed, there does not seem to be any reason why health economists could not inform decision makers with cost utility analyses using both QALYs calculated with patient preferences and those calculated with general public preferences.

4.3. Implementation and its consequences

Drummond et al. (2015) write that: “... in practice, the source of values is often determined as a result of the choice of preference-based instrument” (p.166) (Drummond et al., 2015). Indeed, the key practical barrier to implementation is that an experienced based value set for a generic quality of life instrument such as EQ-5D would have to be developed to have a uniform assessment of benefit across diseases. Since EQ-5D health states are generic descriptions of health, an experience-based value set would require that the person is actually ‘in’ the health state under valuation and recognize it as such. This experienced-based value set could be developed using similar techniques as used for the estimation of the general public value set, as described in a detailed valuation protocol (Oppe et al., 2014). It would require the inclusion of patients who are in one of the 86 health states outlined in the valuation protocol. Hence, in such a setting ‘experience-based’ means ‘experience with the health state under valuation’, not ‘experience with the specific disease causing one to be in a health state’. Alternatives are also feasible: an experience-based value set for EQ-5D has been generated before using TTO in Sweden (Burström et al., 2014), be it with a different approach than detailed in the EuroQoL valuation protocol. Once such a value set has been developed, health economic models simply need to run a scenario analysis using the experienced based EQ-5D value set rather than the general public value set. A consequence of this approach is that it could complicate decision-making in case of conflicting outcomes, as exemplified in scenarios 3 and 4 of Table 3.

In scenarios 1 and 2 in Table 3 the application of two different sources of preference values results in the same outcome in the decision-context: the ICER is either above or below the threshold. Scenarios 3 and 4 pose a challenge as they lead to opposite conclusions. Note that such conflicting scenarios currently also exist, but simply go unnoticed due to the use of only one source of preference values. When explicated, these conflicting results may be explicitly addressed in the appraisal phase. Scenario 4 reflects the situation in which it is in the interest of patients to use general public values. When balancing information from the two perspectives in scenario 4, decision-makers face the question what drives the observed difference. If this is due to adaptation, it may be relevant to know the type of adaptation. For example, if an individual is born with a condition such as cystic fibrosis, decision-makers may conclude that the perspective of the patient is ill-informed due to perception bias: the patient has never been familiar with full health. The decision, then, could sooner be to use general public values than when the adaptation is considered

Table 3

Scenarios for ICERs based on two sources of preference.

	Scenario 1	Scenario 2	Scenario 3	Scenario 4
ICER based on general public preferences	Above threshold	Below threshold	Above threshold	Below threshold
ICER based on patient preferences	Above threshold	Below threshold	Below threshold	Above threshold

‘unbiased’ and patients have simply “learned to live with adversity, and to smile courageously in the face of it” (Cohen, 1993). Then, it may be considered more appropriate to include patient values in societal decisions regarding resource allocations. Scenario 3 may occur in case of life extending interventions and makes clear to policy makers what the value is patients themselves attach to prolonged life. This may lead to more emphasis on life prolonging interventions relative to quality of life improving interventions. While this approach may arguably complicate decision-making, it does give more relevant information to decision makers.

5. Conclusion

Preferences for health states are commonly derived from the general public, rather than from actual patients. We argued that both viewpoints are reasonable and compelling, and that, therefore, **the challenge is to investigate possibilities to intelligently combine the different sources of information.**

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