Nothing About Us, Without Us? A call for involving children in the valuation of health states for QALY estimation

Running title: Involving children in the valuation of health states

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Abstract

Objectives: In many countries, guidelines suggest using EQ-5D instruments for calculating quality-adjusted life years (QALYs) and eliciting adults' preferences to derive utilities. The development of the EQ-5D-Y has given rise to discussion of whose preferences to elicit and whether to (also) elicit preferences of children. Since similar discussions on the involvement of the public have been ongoing in other areas of healthcare, our aim was to (i) provide an overview of arguments for public involvement in decisions on health(care) and (ii) explore to what extent these arguments may hold for involving children in any way in health state valuation (HSV).

Methods: We review theoretical and empirical arguments for public involvement in health(care) and in HSV and map these arguments to the potential involvement of children in HSV.

Results: Several arguments for public involvement can be mapped to the involvement of children in HSV. Examples are involvement being 'a good in itself', it being 'in line with the principles of democracy', and 'consumers having the right to have their voice heard', which are seen as relevant both for public involvement and relevant for involving children in HSV. Empirical arguments furthermore suggest that adults seem to prefer the involvement of children in HSV.

Discussion: Our results indicate that there are several, to some degree competing, arguments that justify the further exploration of involving children in HSV. Additional research is needed to answer remaining questions about the feasibility of and necessary conditions for children's involvement and to provide insight into how to achieve it.

1. Introduction

Cost-utility analyses (CUAs) are increasingly used to inform reimbursement decisions on (new) health interventions (1). In CUAs, the incremental costs and health gains of interventions are compared to relevant comparators like standard care. Health gains are commonly expressed in terms of quality-adjusted life years (QALYs) that combine gains in life duration and health-related quality of life (HRQoL) into a single outcome measure. The utility of HRQoL is typically derived using health state valuation (HSV) tasks in which preferences of adult members of the public are elicited by, for example, asking them to trade-off different life durations for improved health. These utilities are anchored such that 0 represents “dead" and 1 “perfect health” (2). Often, the EQ-5D instrument is recommended for the derivation of utilities for estimating HRQoL gains in patients (3).

Preferences elicited by means of HSV tasks may (albeit indirectly) affect reimbursement decisions by using them for QALY estimations in CUAs. As such, a key normative question that has given rise to ongoing discussions is whose preferences should be elicited, and what their role is in society (4–9). For example, should the preferences be elicited from a (relatively healthy) sample of the general public, as is commonly recommended, or should they be elicited from patients who experience(d) living in impaired health states and will likely utilize the interventions assessed in CUAs?

Discussions about whose preferences should be elicited have become more focal in the context of HSV for HRQoL gains in younger populations (10). Following the increased use of CUAs to inform reimbursement decisions on interventions for paediatric patient populations (11), the EQ-5D was adapted to the EQ-5D-Y to enable the estimation of HRQoL gains in children aged 8–15 years (12). Similar to the valuation protocol for the most recent version of the EQ-5D (the EQ-5D-5L) (used for adults) (13), the current EQ-5D-Y-3L protocol suggests that the utilities of respective health states should be derived from the preferences of adult members of the public by asking them to complete HSV tasks, while 'considering their views about a 10-year-old child' (often referred to as 'child perspective') (12).

The authors of the EQ-5D-Y-3L valuation protocol list three arguments for eliciting preferences from adults rather than from children. Firstly, they argue that it may be considered inappropriate to allow a population in the age range of 8–15 years to decide about matters that concern population health. Secondly, time trade-off (TTO) tasks used for anchoring utilities involve considerations of death and may, therefore, be considered unethical for children (12). Yet, several countries have already made decisions that allow children to be involved in decisions that require such considerations, in the extreme case even when it comes to decisions on actively ending one's life. In the Netherlands, euthanasia is possible for children from the age of 12 with permission of their parents or legal guardians (14). Thirdly, the authors of the valuation protocol posit that it is fair to elicit preferences from adult members of the public as they are taxpayers and ultimately the ones who fund healthcare (12).

The publication of the EQ-5D-Y-3L valuation protocol was followed by empirical work that explored the validity and consequences of eliciting adult preferences to obtain utilities for child HRQoL. Evidence, for example, indicates that adults' completion of HSV tasks from a child perspective yields utilities for EQ-5D-Y-3L health states that seemingly differ from corresponding EQ-5D health states valued by adults for themselves (often referred to as 'adult perspective') (15–18). As discussed by Devlin et al. (19), such perspective-dependent differences need not be a problem as long as the derived utilities are valid and accurately reflect the utility of child HRQoL (vis-à-vis adult HRQoL). Yet, if any such differences stem from considerations and preferences adults may have about aspects of the life of a (10-year-old) child that are not part of the EQ-5D-Y descriptive system, and hence are not included in HSV tasks, such perspective-dependent differences could be cause for concern (as their influence on the derived utilities may not be intended). Such considerations may, for instance, be related to differences in time preferences or result from differences in other types of choice behaviour between the adult and child perspectives (18,20–22). Moreover, while completing HSV tasks for a child, adults have expressed fear of underestimating what it is like for a child to live in impaired health states, as well as feelings of discomfort stating their preferences without knowing the preferences of the child themself (23). Adults have also expressed reluctance to trade off (i.e., give up) life duration for a child (23,24). The decision to exclusively rely on adult preferences for valuing child HRQoL may also conflict with the wishes of adults, in the role of taxpayers, themselves. That is, adults valuing health states from a child perspective question their moral right to state their preferences for a child (23,24), and explicitly state that they believed that children should, and could in some way, be involved in the valuation process (23,25).

Recent stakeholder engagement has indicated that they call for the involvement of adolescents (26,27), and even younger children (in some way appropriate for their age) in HSV (25). Such involvement would be in line with the broader trend of involving members of the public and, more specifically, involving children in (policy) decisions that ‘affect their lives’ (28): nothing about us, without us. For example, national and local (health) authorities increasingly support the participation of children in policy development and implement schemes that foster their involvement in decision making (29–34). In addition, many countries have already been established legal frameworks explicating under which circumstances and from what age children should (also) have a say in decisions on their health (35–37), and it has even been said that being involved is a human right children are entitled to (38). These frameworks demonstrate that (western) societies attach value to children’s views about their health(care) at all ages and from ages of 12 and upwards it is considered important to allow children to state their preferences (i.e., consent) for treatments that influence their own health(care). Seeing as completing HSV tasks also involves stating preferences for health states (albeit hypothetical ones), these frameworks may provide an additional argument for including children's views and potentially even eliciting their preferences from age 12 upwards (with some degree of adult, or parental, involvement). While the hypothetical nature of the tasks complicates the comparison, not involving children in HSV seemingly contrasts a bigger trend of involving children in decisions made in other areas of health(care). Whereas involvement of children in HSV is a topic that surfaced in recent discussions, this broader involvement and exploration of who to involve and why to involve them has been a topic of extensive study and discussion. Some arguments for involvement seem to be mentioned in both discussions, but the extent to which the broader arguments for public involvement also are relevant for and apply to children's involvement in HSV currently remains unclear. Such exploration may be useful as it can enrich the discussion around children's involvement through mapping the arguments from the broader literature on public involvement (39). Therefore, the aim of this paper is to explore which of the broader arguments for public involvement may also apply to involving children in HSV and provide a research agenda to further explore whether and, if so, how children should and could be involved in HSV. Note that we use a broad definition of 'involvement' in this paper as this can encompass various possible approaches that depend on the age and abilities of the children involved, as well as on the role they have. For instance, involving children in HSV can range from them completing HSV tasks themselves (instead of, or in addition to, completion by adults) to policymakers consulting children at some point in the decision-making process on the reimbursement of health interventions for paediatric patient populations. Also note that we primarily discuss arguments *for* involving children in HSV in this paper. We would like to refer the interested reader to Rowen et al. (39) for a discussion of the arguments for not involving them and the challenges associated with their involvement.

To meet our aim, we first discuss the broader arguments posed in the literature for involving members of the public in decisions on health(care). Secondly, we focus on arguments for public and patient involvement in HSV. Finally, we explore to what extent these arguments can be mapped to children’s involvement in HSV and provide a research agenda.

2. Broader arguments for public involvement in decisions about health(care)

The relevance of public involvement in decisions on health(care) has been widely debated and researched (40–46), and its implementations have been ongoing for some time. Already in 1978, the World Health Organization (WHO) stated that "people have a right and duty to participate individually and collectively in the planning and implementation of their health care" (47). Since then, public involvement seems to have gained the attention of several health authorities (34,48,49).

The arguments for public involvement are extensive, considering that it can serve many aims and be implemented on different levels of decision making and in various ways, ranging from the public being involved as mere consultants to policymakers to them having actual decision power (42,43,50,51). In relation to decisions on health(care), an often-made distinction is the role of representative of the general public and the role of patient. While 'public involvement' usually relates to decisions that affect the health and access to healthcare of others in society, 'patient involvement' relates to decisions that directly affect one's own health and access to healthcare (52).

We classify the arguments for public involvement in decisions about health(care) into input, process, and outcome arguments. In doing so, we follow the example of Conklin et al. (2010) who classified the benefits of public involvement in healthcare policy into intrinsic, instrumental, and developmental benefits (45). Below we explain our classification and present several examples of input, process, and outcome arguments.

Table 1 presents an overview of these arguments (numbered 1 to 13 for clarity). Arguments 1 to 4 are input arguments, which are based on the principles underlying public involvement. Arguments 1 to 3 express how public involvement aligns with democratic values. They successively describe how public involvement is considered 'a good in itself' that is 'fitting for democratic societies' since it allows for the voices and preferences of members of the public to be heard, and how consumers of healthcare should have a say in decisions that concern their health (44,45,53,54). Argument 4 is another widely taken perspective on public involvement; taxpayers should serve as decision-makers (12). The public, being crucial financial contributors to the public sector, should have some say in policy decisions – ultimately to ensure (better) alignment of healthcare policy with their preferences and priorities (45,54). They are the payers and potential consumers of healthcare, which may give them a rightful claim to involvement (45,54).

Arguments 5 to 8 are process arguments, which argue for the benefits of public involvement in decision-making. They describe political and educative effects on the individuals involved like, for example, how public involvement can improve the public's understanding of policy (constraints) and can increase the acceptance of changes in policy (44,45,54). Furthermore, being involved increases confidence for (political) engagement and, in the case of individual decision-making, improves understanding of a patient's condition and care (44,45).

Arguments 9 to 13 are outcome arguments, which describe the beneficial consequences of public involvement in decision-making. Argument 9 states that public involvement leads to improved decision-making, achieved due to new, relevant information being included (44,45) and the introduction of lived experiences into policy (41). Furthermore, arguments 10 shows how public involvement can increase the legitimacy of decision outcomes (45) and argument 11 describes improved quality of decisions due to their alignment with the values and priorities of the public (54). Argument 12 describes how some even argue that involving the public in decision making is the best method to achieve the optimal decision outcome for society (4,7). The final argument, argument 13, describes the potential boost of a healthcare system's responsiveness to the needs of healthcare users due to its alignment with society's preferences (45).

For each of these arguments for public involvement in decisions on health(care), a counterargument could potentially be raised, and any involvement will likely go hand in hand with several risks and challenges. Some of these challenges are specific to involving members of the public; however, often they apply to involving any stakeholder with a specific role and interest in the outcome of the decision. For example, a potential risk of public involvement is the subjectivity (i.e., self-interest) of members of the public (53,55). Potential challenges are insufficient resources (e.g., in terms of experiences and knowledge) of those involved (53), increased complexity and duration of the decision-making process (54), and sensitivity to political lobbying of members of the public (53). Bruni et al. (54), however, argue that these challenges hold true for every stakeholder involved in decision-making, including policymakers, physicians, and researchers, and that there is thus no reason to assume that the public would be more subjective than others involved in the decision-making process (54).

Table 1. Broader arguments for public involvement in decisions about health(care)

Classification of arguments	***#***	***The public should be involved, because…***
Input	(1)	public involvement is a good in itself
	(2)	public involvement is fitting in a democratic society
	(3)	consumers should have a say in decisions concerning them
	(4)	taxpayers should have a say in decisions concerning them
Process	(5)	public involvement can improve its understanding of (constraints on) policy
	(6)	public involvement can increase its acceptance of changes in policy
	(7)	public involvement can increase confidence for (political) engagement for the ones involved
	(8)	public involvement can increase its understanding of individual patient's condition and care needs
Outcome	(9)	public involvement leads to the introduction of new and relevant expertise into policy
	(10)	public involvement increases the legitimacy of outcome of decision-making
	(11)	public involvement increases the quality of decisions by ensuring better alignment with public's values and priorities
	(12)	public involvement is the best method to achieve optimal societal decision outcome
	(13)	public involvement can result in a boost to a healthcare system's responsiveness

3. Arguments for public involvement in health state valuation

Table 2 lists the arguments used for public or patient involvement in HSV. It seems that, in the literature on HSV arguments for public involvement are often related to input argument 4. It is, for example, argued in the valuation protocol that the public consists of taxpayers that ultimately fund healthcare (at least in publicly financed healthcare systems) (12). Additionally, it is argued that HSV is tied to the distribution of healthcare resources and, therefore, the public's preferences and involvement are essential (4,12).

In the literature on HSV, other arguments for involvement are role specific and a clear distinction is made between the roles of 1) adult members as representatives of the general public, and 2) patients as representative of those affected by a decision (4–7). There are ongoing discussions on whose preferences should count in HSV (4–9), as it is questioned which of the two roles (public or patient) would be optimal (5–7,56–58).

The arguments posed for public or patient involvement in HSV seem to be predominantly outcome related. Arguments 14 to 16 argue for the involvement of the public rather than the patient, as the public's preferences are considered most suitable. For example, argument 14 describes that 'focusing effects' may cause patients to have recall difficulties, leading them to put too much emphasis on parts (i.e., specific dimensions) of their health state only (4). As compared to patients, members of the public are generally healthy and, therefore, believed to be better able to compare impaired health to full health according to argument 15 (4,59). Argument 16 entails that patients adapt to their health state and, therefore, may underestimate the utility of their own HRQoL (ref to be inserted). However, the validity and accuracy of the public's preferences has also been questioned, as they may be under the influence of similar biases, as illustrated by arguments 17 to 19. For example, members of the public may be less able to value impaired health states, due to 'focusing effects' that may cause members of the public to disproportionally focus on negative aspects of a health state (4,9,56). Other potential biases are ‘valuation compression’ that may cause the general public to struggle with distinguishing between small differences between health states, and ‘contrast effects’ that may cause the general public to underestimate the positive, ‘forgiving effect’ an impaired health state has on other issues in a patient’s life (4). On the other hand, arguments 20 and 21 attach considerable weight to patients’ knowledge and lived experience with regard to (impaired) health and healthcare. In relation to HSV, it is considered relevant that patients adapt to their impaired health state, and therefore it is argued that their adaptation to a health state gives their preferences superiority over those of members of the public, as the public has insufficient information to assess and value (the consequences of) illness (4–7,58). There are also indications that patients are better at imagining dimensions like anxiety and depression, and pain or discomfort than members of the public (8). Argument 22 finds its origin in welfare economics and argues for patient involvement on grounds of well-being in a society being the aggregation of utility of its individual members, therefore attaching considerable importance to the preferences of the ones affected (4).

The arguments distilled from HSV literature seem to be based on different classifications relative to public involvement in healthcare in general. Input arguments (e.g., arguments based on underlying principles of involvement) do not appear to be widely represented in the literature on HSV, with the exception of the taxpayer argument, which seems to serve as the principal argument to elicit utilities from adult members of the public (12). Process arguments (e.g., the process of being involved being beneficial to the public/patients) are not vastly present in the literature either—none of the arguments for public involvement mentioned in the literature on HSV seems to focus on the benefits to the individuals that are involved in the valuation of HRQoL. Most of the arguments in this section, like the focusing effects, the contrast effects, and the adaptation by patients are outcome arguments that focus on the impact of public or patient involvement on the validity and accuracy of the derived HRQoL utilities.

Table 2. Arguments for public or patient involvement in health state valuation

Classification of arguments	***#***	***The public or patients should be involved, because…***	***Argues for…***
Input	(4)	taxpayers should have a say in decisions that concern them	
Outcome	(14)	‘focusing effects’ cause patients to suffer from recall bias, so public preferences are optimal	*public preferences*
	(15)	public is generally healthy and therefore better at imagining full health, so public preferences are optimal	
	(16)	‘adaptation’ to impaired health states by patients leads them to underestimate their HRQoL, so public preferences are optimal	
	(17)	‘focusing effects’ cause the public to emphasize negative aspects of a health state, so patient preferences are optimal	*patient preferences*
	(18)	‘valuation compression’ means that the public is unable to distinguish small differences between health states, so patient preferences are optimal	
	(19)	‘contrast effects’ cause public to disregard minor health issues, so patient preferences are optimal	
	(20)	‘adaptation’ by patients is underestimated by the public, so patient preferences are optimal	
	(21)	patients are better at imagining dimensions like anxiety and depression, and pain and discomfort, so patient preferences are optimal	
	(22)	the well-being of a society is the aggregation of individual utility, so the preferences of the ones affected are vital	

4. Mapping the arguments for public involvement to children

Table 4 describes to what extent arguments 1 to 13 can be mapped from public involvement in health(care) to public or patient involvement in HSV, and whether and how arguments 1 to 22 can be mapped to the involvement of children in HSV. In Table 3 we present the underlying rationale for (not) mapping the arguments based on our own reasoning of whether the argument could apply to the context in question.

The Table shows that input arguments 1 to 3 can be mapped to both public or patient involvement in HSV, as well as to children's involvement, as the arguments (e.g., involvement being fitting in a democratic society) may remain true for public/patient involvement in HSV and when discussing involving children, as they too are part of a democratic society based on which it can be argued that their preferences should be valued. Though democracy means that state power is vested in the general population of societies, which many consider to be adults, it is worth exploring how children can enrichen democratic decision-making with their preferences and expertise on recent lived experiences. Argument 4 is presented as an important argument when it comes to public involvement in health(care) and in HSV, but it does not map to the involvement of children, as the majority of children may not yet pay any taxes – which may even be used as an argument against involving them in HSV (60). However, older children with a job may pay taxes (61), in which case the argument would hold for those children.

Of process arguments 5 to 8, number 8 is the only one that maps to the involvement of children. Here, arguments like involvement resulting in improved understanding of constraints on policy, are not mentioned in HSV literature and are hard to argue for when it comes to the involvement of children. While children could learn from the process of being involved, it seems unlikely that their understanding of policy constraints meaningfully improves (ref to be inserted).

Outcome arguments 9 to 12 all map to from public involvement in health(care) to the involvement of children. Firstly, it is argued that it leads to potential improvement of decision making and an increased legitimacy of the decisions made (ref to be inserted). Involving children in decisions that concern them, may align decisions (and their legitimacy) with their preferences—depending on how children are involved and what value is attached to their preferences and opinions. Furthermore, the involvement of children may bring new expertise to the table that is unique to their point of view and cannot easily be reproduced by others (who are no longer children themselves) (26). Argument 13 is the only outcome argument that does not map to the involvement of children.

Outcome arguments 14 to 22, originating from public/patient involvement in HSV all map to the involvement of children in HSV. The various arguments made for the use of public preferences rather than patient preferences may not be easily translated to the involvement of children, though they do not provide a basis on which to argue against it. Patient-based arguments are more easily mapped to the involvement of children. That is, if significant weight is attached to the preferences of patients as experts based on the assumption that this increases accuracy of HSV, similarly, weight should be attached to the preferences of children who are patients, as they can therefore contribute relevant expertise and lived experience.

Table 3. Overview and mapping of arguments per layer, including classification of the type of arguments.

Health(care)	***Health state valuation***	***Children's involvement***	***Explanation of reasoning behind mapping***
(1)[1] →	→	→	*Public involvement being 'a good in itself' remains true when talking about public/patient involvement in HSV and for children's involvement.*
(2)[1] →	→	→	*Public involvement being being fitting in a democratic society remains true when talking about public/patient involvement in HSV and for children's involvement.*
(3)[1] →	→	→	*Consumers should have a say in decisions concerning them, which maps to the involvement of both adult and child patients in HSV.*
(4)[1] →	→	▨	*The taxpayer argument is the primary argument for public involvement in HSV. It cannot be mapped to children's involvement, since (most) children do not pay taxes.*
(5)[2]	▨	▨	*The argument of public involvement improving the understanding of (constraints on) policy is not used in HSV literature. It might be true that children could learn from being involved, which could be an argument for their involvement. However, we are unaware of relevant evidence showing this effect in child HSV. Therefore, this argument does not map to children's involvement.*
(6)[2]	▨	▨	*The argument of public involvement increasing acceptance of changes in policy is not used in HSV literature. Furthermore, it is unlikely that children are aware enough of changes in policy regarding HSV.*
(7)[2]	▨	▨	*The argument of public involvement increasing confidence for (political) engagement is not used in HSV literature. Furthermore, we are unaware of relevant evidence showing this effect in child HSV. Therefore, this argument does not map to children's involvement.*
(8)[2] ⇢	▨ ⇢	⇢	*The argument of public involvement increasing the understanding of individual patient's condition and care is not used in HSV literature. It might, however, be used to argue for the involvement of children in HSV, as it potentially increases the understanding of the children that are involved about their own health(care).*
(9)[3] →	→	→	*Public and/or patient involvement in HSV leads to the introduction of new and relevant expertise into policy, which also remains true for children's involvement.*
(10)[3] →	→	→	*Public and/or patient involvement in HSV may increase the legitimacy of outcome of decision-making, which also remains true for children's involvement.*
(11)[3] →	→	→	*Public and/or patient involvement may increase the quality of decisions by ensuring better alignment with the public's or patient's values and priorities, which also remains true for children's involvement.*
(12)[3] →	→	→	*The argument that public involvement is the best method to achieve optimal societal decision outcome can be used to argue both for public/patient involvement in HSV and for children's involvement.*

(13)[3]			*Value sets, due to their indirect contribution to healthcare decision-making are unlikely to improve the responsiveness of healthcare systems in any meaningful way.*
	(14)[3]	→	*'Focusing effects' cause patients to suffer from recall bias, which calls for relying on public preferences. There is insufficient evidence on the effect of this bias on children in HSV. However, as children can fulfil the role of members of the general public, there is no reason to argue against their involvement based on this argument.*
	(15)[3]	→	*The public is generally healthy and therefore better at imagining full health, so public preferences are optimal. As children can fulfil the role of members of the general public, there is no reason to argue against their involvement based on this argument.*
	(16)[3]	→	*'Adaptation' by patients leads them to underestimate their HRQoL, so public preferences are optimal. As children can fulfil the role of members of the general public, there is no reason to argue against their involvement based on this argument.*
	(17)[3]	→	*'Focusing effects' cause the public to emphasize negative aspects of a health state, so patient preferences are optimal. Based on this argument, child patients should be involved in HSV.*
	(18)[3]	→	*'Valuation compression': public is unable to distinguish small differences between health states, so patient preferences are optimal. Based on this argument, child patients should be involved in HSV.*
	(19)[3]	→	*'Contrast effects' cause public to disregard minor health issues, so patient preferences are optimal. Based on this argument, child patients should be involved in HSV.*
	(20)[3]	→	*'Adaptation' by patients is underestimated by the public, so patient preferences are optimal. Based on this argument, child patients should be involved in HSV.*
	(21)[3]	→	*Patients are better at imagining dimensions like anxiety and depression, and pain and discomfort, so patient preferences are optimal. Based on this argument, child patients should be involved in HSV.*
	(22)[3]	→	*Well-being of a society is the aggregation of individual utility, so the accuracy of children's utility could be crucial. If involving children improves accuracy, this argument applies.*
Input, process, outcome	***Predominantly outcome related***		

The arrows illustrate whether and how the arguments can be mapped to the other layers.

[1]Input arguments, [2]Process arguments, [3]Outcome arguments

5. Discussion

The aim of this study was to explore which of the broader arguments for public involvement may also apply to involving children in HSV and provide a research agenda to further explore whether and, if so, how children should and could be involved in HSV.

It seems as if the decision to involve the general public or patients relies on two (potentially competing) sets of arguments: 1) (input) arguments that are based on the underlying principles of public involvement (e.g., the taxpayer perspective and arguments based on the values of a society, like involvement being an integral part of a democracy), and 2) (outcome) arguments that are based on the (assumed) accuracy and validity of the utilities elicited in HSV tasks (i.e., whose preferences perform best). The sets of arguments seem to compete with one another as they are based on different viewpoints on what should matter—do we want to make sure that the methods chosen for HSV align with the underlying principles we deem important, or do we want to optimize the accurateness of the outcomes of the chosen methods? If and how we involve children, depends on which of the sets of arguments is considered most essential. However, both of these sets of arguments provide reason to explore the involvement of children. If the underlying principles of the decision-making process are what we want to focus on (i.e., input arguments matter most), children's preferences can be considered important on the basis of, for example, their right to have a say. Additionally, if the objective is to ensure accuracy and validity of utilities in HSV (i.e., output arguments matter most), children could play a role to increase alignment with their experiences and views of HRQoL and to minimize potential biases stemming from adults valuing child health states.

In this paper, we explored the arguments for involving children in HSV by examining literature discussing similar discussions on other levels of healthcare and ultimately, we call for children involvement. Nevertheless, some limitations need to be discussed. First of all, due to the focus on HSV, this paper mostly discusses literature on health economics and outcomes research and therefore we likely missed valuable insights from other fields that could contribute to the discussion. Moreover, the arguments mentioned in this paper are mainly in favour of public involvement. Though some challenges of the implementation of involving the public and arguments against doing so are presented, the literature that we rely upon largely focuses on the benefits. On the level of public involvement in healthcare in general, this might be because consensus seems to have been reached on its importance, which is highlighted by authorities' desire to involve the public. As we use a trickle-down approach that starts from the macro-level of healthcare in general, this focus persists throughout the paper. Also, the aim of this paper was to investigate whether it is desirable to explore the possibilities of involving children in some, as of yet undefined, way. Naturally, when looking solely at involving children in HSV (i.e., without a mapping approach), arguments against their involvement become more prevalent.

Research agenda

Further research should focus on several matters. As this article distils two competing sets of arguments, one of which focuses on the (assumed) accuracy and validity of the utilities elicited in HSV tasks. This should be further explored. That is, if the utilities elicited for children through particular instruments perform well when it comes to accuracy and validity, the sets of arguments are no longer competing, and that would shift the focus of the discussion on children's involvement.

Moreover, it should be examined what the views of policymakers working on reimbursement decisions are regarding the arguments posed in this paper. They are the ones that ultimately decide, and their opinions on the involvement of children in their decisions is crucial. Secondly, a similar examination should be carried out with members of the public (that is, children, adolescents, and adults).

After these examinations have provided a clear overview of the potential desirability of involving children in HSV, ways in which children could be involved (with or without directly eliciting their preferences) should be explored (e.g., through consultations with stakeholders, systematic reviews of methods for children involvement, as well as head-to-head comparisons of specific methods).

Conclusions

The mapping exercise performed in this paper sheds light on lessons that can be learned from discussions on public involvement (of children). While it might be difficult to determine what role children could and should play, we see ample reason to explore the feasibility of children involvement in HSV: nothing about us, without us. Nevertheless, questions remain concerning the feasibility of involving children in HSV, that can be explored in future research.

References

1. Kwon J, Kim SW, Ungar WJ, Tsiplova K, Madan J, Petrou S. Patterns, trends and methodological associations in the measurement and valuation of childhood health utilities. Quality of Life Research. 2019 Jul 15;28(7):1705–24.
2. Pliskin JS, Shepard DS, Weinstein MC. Utility functions for life years and health status. Oper Res. 1980;206–24.
3. Kennedy-Martin M, Slaap B, Herdman M, van Reenen M, Kennedy-Martin T, Greiner W, et al. Which multi-attribute utility instruments are recommended for use in cost-utility analysis? A review of national health technology assessment (HTA) guidelines. The European Journal of Health Economics. 2020 Nov 8;21(8):1245–57.
4. Helgesson G, Ernstsson O, Åström M, Burström K. Whom should we ask? A systematic literature review of the arguments regarding the most accurate source of information for valuation of health states. Quality of Life Research. 2020 Jun 1;29(6):1465–82.
5. Versteegh MM, Brouwer WBF. Patient and general public preferences for health states: A call to reconsider current guidelines. Vol. 165, Social Science and Medicine. Elsevier Ltd; 2016. p. 66–74.
6. Brazier J, Akehurst R, Brennan A, Dolan P, Claxton K, Mccabe C, et al. Should Patients Have a Greater Role in Valuing Health States? Vol. 4, Appl Health Econ Health Policy. 2005.
7. Brazier J, Rowen D, Karimi M, Peasgood T, Tsuchiya A, Ratcliffe J. Experience-based utility and own health state valuation for a health state classification system: why and how to do it. European Journal of Health Economics. 2018 Jul 1;19(6):881–91.
8. Ogorevc M, Murovec N, Fernandez NB, Rupel VP. Questioning the differences between general public vs. patient based preferences towards EQ-5D-5L defined hypothetical health states. Health Policy (New York). 2019 Feb 1;123(2):166–72.
9. Peeters Y, Vliet Vlieland TPM, Stiggelbout AM. Focusing illusion, adaptation and EQ-5D health state descriptions: The difference between patients and public. Health Expectations. 2012 Dec;15(4):367–78.
10. Rowen D, Rivero-Arias O, Devlin N, Ratcliffe J. Review of Valuation Methods of Preference-Based Measures of Health for Economic Evaluation in Child and Adolescent Populations: Where are We Now and Where are We Going? Pharmacoeconomics. 2020 Apr 6;38(4):325–40.
11. Sullivan SM, Tsiplova K, Ungar WJ. A scoping review of pediatric economic evaluation 1980-2014: do trends over time reflect changing priorities in evaluation methods and childhood disease? Expert Rev Pharmacoecon Outcomes Res. 2016 Sep 2;16(5):599–607.
12. Ramos-Goñi JM, Oppe M, Stolk E, Shah K, Kreimeier S, Rivero-Arias O, et al. International Valuation Protocol for the EQ-5D-Y-3L. Pharmacoeconomics. 2020 Jul 16;38(7):653–63.
13. Stolk E, Ludwig K, Rand K, van Hout B, Ramos-Goñi JM. Overview, Update, and Lessons Learned From the International EQ-5D-5L Valuation Work: Version 2 of the EQ-5D-5L Valuation Protocol. Value in Health. 2019 Jan 1;22(1):23–30.

14. Rijksoverheid. Wie kan om euthanasie vragen? [Internet]. [cited 2024 May 8]. Available from: https://www.rijksoverheid.nl/onderwerpen/levenseinde-en-euthanasie/vraag-en-antwoord/wie-kan-om-euthanasie-vragen

15. Kreimeier S, Oppe M, Ramos-Goñi JM, Cole A, Devlin N, Herdman M, et al. Valuation of EuroQol Five-Dimensional Questionnaire, Youth Version (EQ-5D-Y) and EuroQol Five-Dimensional Questionnaire, Three-Level Version (EQ-5D-3L) Health States: The Impact of Wording and Perspective. Value in Health. 2018 Nov;21(11):1291–8.

16. Shah KK, Ramos-Goñi JM, Kreimeier S, Devlin NJ. An exploration of methods for obtaining 0 = dead anchors for latent scale EQ-5D-Y values. The European Journal of Health Economics. 2020 Sep 6;21(7):1091–103.

17. Lipman SA, Reckers-Droog VT, Karimi M, Jakubczyk M, Attema AE. Self vs. other, child vs. adult. An experimental comparison of valuation perspectives for valuation of EQ-5D-Y-3L health states. European Journal of Health Economics. 2021 Dec 1;22(9):1507–18.

18. Lipman SA, Zhang L, Shah KK, Attema AE. Time and lexicographic preferences in the valuation of EQ-5D-Y with time trade-off methodology. The European Journal of Health Economics. 2023 Mar 21;24(2):293–305.

19. Devlin NJ, Pan T, Sculpher M, Jit M, Stolk E, Rowen D, et al. Using Age-Specific Values for Pediatric HRQoL in Cost-Effectiveness Analysis: Is There a Problem to Be Solved? If So, How? Pharmacoeconomics. 2023 Oct 13;41(10):1165–74.

20. Lang Z, Attema AE, Lipman SA. The effect of duration and time preference on the gap between adult and child health state valuations in time trade-off. European Journal of Health Economics. 2023;

21. Hoogenboom AFH, Lipman SA. Loss aversion in EQ-5D-Y-3L: does it explain differences in willingness to trade-off life duration in adult and child perspectives? EsCHER Working Paper Series No 2024002. 2024;

22. Lipman SA, Reckers-Droog VT. Comparing heuristic valuation processes between health state valuation from child and adult perspectives. The European Journal of Health Economics. 2024 Feb 3;

23. Reckers-Droog V, Karimi M, Lipman S, Verstraete J. Why Do Adults Value EQ-5D-Y-3L Health States Differently for Themselves Than for Children and Adolescents: A Think-Aloud Study. Value in Health. 2022 Jul;25(7):1174–84.

24. Åström M, Conte H, Berg J, Burström K. ‘Like holding the axe on who should live or not’: adolescents’ and adults’ perceptions of valuing children’s health states using a standardised valuation protocol for the EQ-5D-Y-3L. Quality of Life Research. 2022 Jul 1;31(7):2133–42.

25. Powell PA, Rowen D, Keetharuth A, Mukuria C. Understanding UK public views on normative decisions made to value health-related quality of life in children: A qualitative study. Soc Sci Med. 2024 Jan;340:116506.

26. Nazari JL, Pickard AS, Gu NY. Findings from a Roundtable Discussion with US Stakeholders on Valuation of the EQ-5D-Y-3L. Pharmacoeconomics. 2022 Dec 29;40(S2):139–46.

27. Xie F, Xie S, Pullenayegum E, Ohinmaa A. Understanding Canadian stakeholders’ views on measuring and valuing health for children and adolescents: a qualitative study. Quality of Life Research. 2024 May 5;33(5):1415–22.

28. Gottschalk F, Borhan H. Child participation in decision making: Implications for education and beyond. 2023 Oct.

29. British Youth Council. About us.

30. NHS England. NHS Youth Forum.

31. Gemeente.nu. Betrek jeugd bij plannen en beleid. 2018.

32. Nederlandse Wetenschapsagenda. Manifest Jongeren Challenge. 2021.

33. Council of Europe. Child Participation. 2024.

34. World Health Organisation. WHO/Europe urges governments to include young people in decisions about their health. 2022.

35. NHS England. Children and young people. 2022.

36. Patiëntenfederatie Nederland. Jouw rechten als patiënt.

37. UNICEF. Convention on the Rights of the Child Text.

38. Kinderombudsman de. Mijn zorg mijn zaak.

39. Rowen D, Mukuria C, Powell PA, Wailoo A. Exploring the Issues of Valuing Child and Adolescent Health States Using a Mixed Sample of Adolescents and Adults. Pharmacoeconomics. 2022 May 16;40(5):479–88.

40. Djellouli N, Jones L, Barratt H, Ramsay AIG, Towndrow S, Oliver S. Involving the public in decision-making about large-scale changes to health services: A scoping review. Health Policy (New York). 2019 Jul;123(7):635–45.

41. Boivin A, Lehoux P, Burgers J, Grol R. What are the key ingredients for effective public involvement in health care improvement and policy decisions? A randomized trial process evaluation. Milbank Quarterly. 2014;92(2):319–50.

42. Charles C, Demaio S. Lay Participation in Health Care Decision Making: A Conceptual Framework [Internet]. 1993. Available from: http://read.dukeupress.edu/jhppl/article-pdf/18/4/881/357692/ddjhppl_18_4_881.pdf

43. Mitton C, Smith N, Peacock S, Evoy B, Abelson J. Public participation in health care priority setting: A scoping review. Vol. 91, Health Policy. 2009. p. 219–28.

44. Broqvist Mari. Asking the public : Citizens ́views on priority setting and resource allocation in democratically governed healthcare. Linköping University Electronic Press; 2018. 147 p.

45. Conklin A, Slote Morris Z, Nolte E. Involving the public in healthcare policy: An update of the research evidence and proposed evaluation framework [Internet]. Available from: www.rand.org

46. van Til JA, Pearce A, Ozdemir S, Hollin IL, Peay HL, Wu AW, et al. Role Preferences in Medical Decision Making: Relevance and Implications for Health Preference Research. The Patient - Patient-Centered Outcomes Research. 2024 Jan 24;17(1):3–12.

47. World Health Organisation. Declaration of Alma-Ata.

48. NHS England. Engaging local people: A guide for local areas developing Sustainability and Transformation Plans. 2016.

49. Government of Canada. The Health Canada Policy Toolkit for Public Involvement in Decision Making. 2000.

50. Morgan LM. Community participation in health: perpetual allure, persistent challenge. Health Policy Plan. 2001 Sep 1;16(3):221–30.

51. Arnstein SR. A Ladder Of Citizen Participation. Journal of the American Planning Association. 1969;35(4):216–24.

52. McCoy MS, Warsh J, Rand L, Parker M, Sheehan M. Patient and public involvement: Two sides of the same coin or different coins altogether? Bioethics. 2019 Jul 8;33(6):708–15.

53. Cleemput I, Christiaens W, Kohn L, Léonard C, Daue F, Denis A. Acceptability and Perceived Benefits and Risks of Public and Patient Involvement in Health Care Policy: A Delphi Survey in Belgian Stakeholders. Value in Health. 2015 Jun 1;18(4):477–83.

54. Bruni RA, Laupacis A, Martin DK. Public engagement in setting priorities in health care. Can Med Assoc J. 2008 Jun 12;179(1):15–8.

55. Litva A, Coast J, Donovan J, Eyles J, Shepherd M, Tacchi J, et al. “The public is too subjective”: public involvement at different levels of health-care decision making. Vol. 54, Social Science & Medicine. 2002.

56. Damschroder LJ, Zikmund-Fisher BJ, Ubel PA. The impact of considering adaptation in health state valuation. Soc Sci Med. 2005 Jul;61(2):267–77.

57. Mann R, Brazier J, Tsuchiya A. A comparison of patient and general population weightings of EQ-5D dimensions. Health Econ. 2009;18(3):363–72.

58. Menzel P, Dolan P, Richardson J, Olsen JA. The role of adaptation to disability and disease in health state valuation: a preliminary normative analysis. Vol. 55, Social Science & Medicine. 2002.

59. Ernstsson O, Burström K, Heintz E, Mølsted Alvesson H. Reporting and valuing one’s own health: a think aloud study using EQ-5D-5L, EQ VAS and a time trade-off question among patients with a chronic condition. Health Qual Life Outcomes. 2020 Dec 1;18(1).

60. Kreimeier S, Greiner W. EQ-5D-Y as a Health-Related Quality of Life Instrument for Children and Adolescents: The Instrument’s Characteristics, Development, Current Use, and Challenges of Developing Its Value Set. Value in Health. 2019 Jan;22(1):31–7.

61. Bailey C, Howell M, Raghunandan R, Salisbury A, Chen G, Coast J, et al. Preference Elicitation Techniques Used in Valuing Children’s Health-Related Quality-of-Life: A Systematic Review. Pharmacoeconomics. 2022 Jul 27;40(7):663–98.