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Understanding UK public views on normative decisions made to value health-related quality of life in children: A qualitative study

Philip A. Powell^{*}, Donna Rowen, Anju Keetharuth, Clara Mukuria

Sheffield Centre for Health and Related Research, University of Sheffield, UK

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ABSTRACT

Developing methodology for measuring and valuing child health-related quality of life (HRQoL) is a priority for health technology agencies. One aspect of this is normative decisions that are made in child HRQoL valuation. This qualitative study aimed to better understand adult public opinion on the normative questions of whose preferences to elicit (adults, children, or both) and from which perspective (who should be imagined living in impaired health), when valuing child HRQoL. Opinions of the adult UK public (N = 32) were solicited using online semi-structured focus groups, featuring a breadth of age, sex, ethnicities, and responsibility for children under 18 years. Participants were provided with bespoke informational material on health state valuation and were probed for their views. Arguments for and against different positions were discussed. Data was analysed using framework analysis. Participants demonstrated near-to-universal agreement that children should be involved in valuation in some form, yet this should differ depending on age or maturity. There was strong support for approaches combining involvement from children and adults (e.g., their parents), especially for younger children. There was little intuitive support for the 'taxpayer argument' for asking taxpaying adults. In the context of greater involvement of children in valuation, most participants supported using an 'own' perspective. Most participants thought that valuation study participants should know the exercise is about valuing child health states for ethical reasons. Informed views from the UK public on who should be asked and with what perspective when valuing child HRQoL appear to differ from normative positions previously advocated by some health economists, such as prioritising the preferences of taxpaying adults. In contrast, the results suggest including adults and children in valuation, with the proviso that the children are of an appropriate age and level of maturity, and that an own perspective is used wherever possible.

1. Introduction

Health technology assessment (HTA) aims to support societal decision-making in healthcare due to the limitations of markets in this context. Information on the cost-effectiveness of interventions, with respect to beneficiaries who governments represents, is therefore required for HTA. A commonly accepted metric to assess effectiveness is the quality-adjusted life year (QALY), which combines information on length of life with health-related quality of life (HRQoL) into a single score. The QALY characterises length of life multiplied by HRQoL, where HRQoL is scored using 'utility' values anchored at 0 (dead) and 1 (full health), with negative values representing states worse than dead (Torrance and Feeny, 1989). Estimates of HRQoL are typically acquired from adult or child preference-based measures (PBMs) or vignettes,

which combine patients' descriptive scores on particular dimensions of HRQoL with a set of societal preference (or value) weights for living in different health states. These preference weights are "off-the-shelf" values, derived in standardised valuation or preference elicitation studies, which are used to generate the utility values described above.

For submission to many HTA agencies utility values reflect the preferences of all potential beneficiaries of healthcare rather than patients *per se*, and have generally been estimated in adult populations based on a 'taxpayer argument' i.e., members of the adult public primarily and collectively bear the costs of healthcare and their views should be prioritised. While guidance is available on the valuation of adult health, there is less of an established position on the valuation of health for children by HTA agencies, where potential beneficiaries (i.e. children) differ from wider society and taxpayers (i.e. adults). The

^{*} Corresponding author. Sheffield Centre for Health and Related Research School of Medicine and Population Health University of Sheffield Regent Court, 30 Regent Street, Sheffield, S1 4DA, UK.

E-mail address: p.a.powell@sheffield.ac.uk (P.A. Powell).

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decision of whose views should be elicited is normative, and therefore different approaches may be adopted globally. For example, the latest National Institute for Health and Care Excellence (NICE) HTA manual states that utility values for HTAs in the UK should be generated from preferences elicited from a representative sample of the public using an established choice based technique (National Institute for Health and Care Excellence (NICE), 2022). While no explicit position is adopted in the manual on what perspective is taken or whether the sample is restricted to adults (rather than adolescents or children), the standard UK approach for valuing adult health is for an adult sample to take an ‘own’ perspective, or imagine themselves living in the described health states (or different levels of HRQoL). NICE does not have an explicit position on valuing health for children, noting that “there are methodological challenges when developing value sets for children and young people”. (National Institute for Health and Care Excellence (NICE), 2022).

When valuing HRQoL in children there are at least two critical and interrelated normative decisions that need to be made (Rowen et al., 2020). The first is who is asked to provide their values: adults or children and adolescents themselves. The second is who they are asked to think about when providing their values (i.e., what perspective they are asked to take): thinking of themselves or another person (e.g., a younger child). Further methodological intricacies become evident as a consequence of each decision. For example, if children are asked to provide their values, what age range and/or choice-based techniques are appropriate? If adults are asked to value HRQoL for children from a child’s perspective, what age child should they be asked to think about? These two normative decisions matter, as different choices have been shown to lead to different values, (Mott et al., 2021; Lipman et al., 2021a) which ultimately affects the calculation of QALYs for HTA and potentially impacts healthcare funding decisions. Accordingly, answers to these questions are necessary to help decision-makers adopt a reasoned and evidenced position on the valuation of child and adolescent health.

As one of the first attempts at a standardised approach to valuing a child and adolescent PBM across different countries, the valuation protocol for the EQ-5D-Y-3L was published in 2020 (Ramos-Goñi et al., 2020). The EQ-5D-Y-3L is a widely-used generic PBM instrument recommended for children aged 8–15 years old (EuroQol Research Foundation, 2020). For valuation of the EQ-5D-Y-3L, the protocol recommended that a sample of the adult general population valued the health states thinking about a 10-year-old child. The former mirrors the traditional approach for valuing adult measures and was justified with reference to a ‘taxpayer argument’. However, no rationale was given for the use of a 10-year-old child perspective and arguments could be levied for and against this position (Lipman et al., 2021b). A number of value sets for different countries have now been produced following the EQ-5D-Y-3L protocol, with more studies ongoing, though notably there is currently no UK value set (Devlin et al., 2022a).

As part of EuroQol Research Foundation funded EQ-5D-Y-3L valuation studies, investigators were encouraged to carry out stakeholder engagement work to help evaluate the valuation process in a country-specific context (Devlin et al., 2022b). One of the first studies to publish their results in the US conducted a roundtable with 14 stakeholders (including researchers, HTA representatives, and two lay people with experience with children). (Nazari et al., 2022) These stakeholders questioned the use of adult-only preferences, with support for the inclusion of adolescents (either exclusively or in addition to adult views). Further, concerns were raised about heterogeneity in participants’ responses through the use of the ‘10-year-old child’ perspective, depending on their respective experience with children (e.g., parental status).

Historically, normative decisions on whom to ask and with what perspective have been made as a result of different arguments proposed by experts, (Helgesson et al., 2020) such as health economists, with little consultation of the general public who are prominent ‘stakeholders’ in the healthcare system. The exclusion of public views in decisions that

ultimately affect them contradicts both the essence of the ‘taxpayer argument’, used to justify the inclusion of adult public values in health state valuation, and patient and public involvement and engagement (PPIE) initiatives in health research (Versteegh and Brouwer, 2016). Indeed, NICE’s own PPIE policy states that children should be involved “on matters pertaining to NICE’s work and that affect children and young people’s health and wellbeing”. (National Institute for Health and Care Excellence (NICE), 2013).

The current work was designed to begin to fill the gap of not adequately including the public’s views in normative and methodological decisions that have implications for healthcare funding. A focus group methodology with members of the UK public was designed to address the research question: ‘What are the opinions of the general public on whose preferences should be elicited and which perspective should be used when valuing the health-related quality of life (HRQoL) of children and young people?’ Similar qualitative studies conducted on complementary normative issues in health state valuation with the UK public have yielded important insights, including that the same health state is viewed differently when adults use a child perspective or imagine themselves living in the state, (Powell et al., 2021) and that members of the public may not intuitively support the use of an uninformed, representative adult general public sample completing adult valuation tasks (Powell et al., 2022). Such insights are likely to be of interest to international researchers and UK policy makers working in the field.

2. Methods

2.1. Design

A protocol was archived online a priori (<https://doi.org/10.15131/shef.data.20424915.v1>). This research used a qualitative focus group design, chosen as it allows for the direct clarification of issues and content with the researcher, which is important in a complex topic. Further, focus groups allow people to discuss and build upon views shared by others, which can help to enhance understanding and engagement (Gill et al., 2008). A framework analysis approach was adopted as it enables a combination of deductive and inductive knowledge production (Gale et al., 2013). Framework analysis combines multiple elements of qualitative research paradigms and has been labelled a ‘whole paradigm approach’. (Kiernan and Hill, 2018).

2.2. Recruitment and participants

Five semi-structured focus groups were conducted with 5–7 participants in each group (N = 32). While recruitment was pre-determined and not based on observed data saturation, three to six focus groups with six to eight participants has been shown to be sufficient to identify over 90% of themes in qualitative data. (Guest et al., 2017), and this informed the planned sample size. Adult members of the English public were sampled purposively across a breadth of age, sex, ethnicity, and whether participants had responsibility for a child(ren) under 18 years old. Participants were recruited by the market research agency MRFRG (www.mrfgr.com) and compensated by standard market rates (£50 for participation in a 90-min focus group).

2.3. Materials

In order to help explain the topic area and research questions to the public, a bespoke animated introductory informational video was produced (https://drive.google.com/file/d/1N2OLiaVO-XbNcd9IXAlnfJuXTfSsdA1x/view?usp=share_link). This was developed in conjunction with a Patient and Public Advisory Group (PAG; see below). The 7-min video was designed in PowerPoint with a series of animated slides and accompanying narration designed to convey in lay terms the research context and normative issues of interest, including health state valuation

and its application in children. A semi-structured topic guide (Supplementary File A) was produced to ensure all important information was covered. Finally, a 10-item background questionnaire (Supplementary File B) was designed to: enable a description of the sample on key sociodemographic characteristics (7-items on age, sex, gender, ethnicity, level of education, geographical location, parent/guardianship of child(ren) under 18); capture quantitative information on level of understanding of the introductory video with a 5-point response scale (very difficult – very easy) with an option for a free-text explanatory response if ‘very difficult’ or ‘fairly difficult’ was selected; and a free-text feedback question. The informational video, topic guide, and background questionnaire were discussed in a group discussion with the PAG prior to data collection. Feedback was overall positive. Several revisions were made to the video (e.g., clarifying meaning of ‘value’, emphasising the ‘so what’ question earlier on), topic guide (e.g., additional prompts on ‘parents’ and age range), and background questionnaire (e.g., revision of question(s) on sex and gender, and inclusion of open-text feedback) as a result of PAG input.

2.4. Procedure

Written informed consent was obtained prior to the focus group and confirmed with participants at the start of the session. Focus groups were held online using videoconferencing software, Google Meet, in August 2022, outside of core working hours, to facilitate participation by working members of the public and people from a wide geographical area. The focus groups were scheduled to last for 90 min and facilitated by an experienced qualitative researcher with co-facilitation support from a trained health economist, with expertise in health state valuation methodologies. No relationship existed between the researchers and participants prior to the focus groups.

The focus groups were audio recorded and had two phases: an informational phase and a discussion phase. In the informational phase, the facilitator explained the research and participants were asked to watch the introductory informational video. Following the video, participant understanding was probed qualitatively and any misunderstandings were clarified. In the discussion phase, a semi-structured discussion was facilitated in three areas: (i) understanding of HRQoL; (ii) who should be asked to value HRQoL for children and adolescents; and (iii) who should people be asked to think about when valuing HRQoL for children and adolescents (which perspective). Ethics approval for this study was obtained from the host institution (reference number: 046269).

2.5. Analysis

Responses to the questionnaire were descriptively summarised. Focus group recordings were transcribed intelligent verbatim (i.e., omitting redundant, repeated, or filler words and sounds), anonymised, and checked for accuracy. Framework analysis was used, (Ritchie and Spencer, 2002) via six stages (Gale et al., 2013).

- (i) Familiarisation. Four members of the research team involved in data collection and/or analysis read the transcripts and listened to the audio recordings.
- (ii) Coding. An a priori coding framework was produced based on the topic guide. Two researchers independently coded the first two transcripts in hard copy. Inductive themes and potential revisions to the framework were identified.
- (iii) Refining the framework. Two researchers met to discuss the initial coding and consensus was reached on revisions to the framework. The lead researcher used the revised framework to code the remaining three transcripts, noting inductive themes and framework revisions. A final meeting between four researchers was held to discuss the coding and to agree a finalised framework.

- (iv) Indexing. The lead researcher applied the finalised framework to fresh copies of the transcripts on NVivo v1.7.
- (v) Charting. Indexed data was charted in a matrix on Microsoft Excel, organised with a separate sheet for each category, one row per theme, one column per focus group participant, and each cell featuring supportive verbatim extracts of text. The indexed data was checked by the wider research team for agreement.
- (vi) Interpretation. A descriptive summary was written for each theme and the framework was mapped diagrammatically. The lead researcher coded text relevant to arguments as either supportive (green), against (red), or neutral (black), relative to the theme under which they were organised. This coded data was checked for agreement by the wider research team.

Trustworthiness of the analysis was assured in four ways.

- (i) Triangulation. The first two transcripts (33%) were dual-coded and meetings were held to discuss the coding and refine the analytic framework within the research team.
- (ii) Peer debriefing with PAG collaborators. An overview of the analysis, findings, exemplary supporting data, and interpretative decisions was presented to the PAG for feedback. PAG collaborators were asked whether they agreed with the themes that emerged from the analysis and whether anything was surprising, unclear, or missing. They were also asked whether and how the presentation of data could be improved.
- (iii) Auditable decision trail and transparency. All methodological and coding decisions were recorded, allowing for a transparent and traceable audit trail from the raw data to the finalised framework.
- (iv) Reflective journal. To enhance reflexivity, the lead researcher kept a reflective journal throughout the research (Ortlipp, 2008). This journal was designed for the primary researcher to document their presuppositions, choices, experiences and actions throughout the qualitative research process.

2.6. Patient and public involvement and engagement

A Patient and Public Advisory Group (PAG) was convened to collaborate on the study. Invitations were distributed to the NICE Public Involvement Programme (PIP) Expert Panel and the Patient and Public Involvement Panel for the Policy Research Unit in Economic Methods of Evaluation of Health and Social Care Interventions (EEPRU), looking to engage 3–4 people from each to work on the study. Expressions of interest were sought from people of a range of ages and genders. Criteria for selection also included at least one person from an ethnic minority background and at least one person who was a parent/guardian for children aged under 18 years old.

Eight people were invited to collaborate on the study (four from the NICE panel and four from the EEPRU panel). Ages of invitees ranged from 22 to 63 and included two men, one person from an ethnic minority background, and two parent/guardians of children aged under 18 years old (other collaborators had older children).

Two meetings of the PAG were convened during the course of the study, first in March 2022 to collaborate and input into study materials, and second in December 2022 to discuss and feedback on the study findings and presentation of data. Four members of the PAG contributed to the first meeting. Six members of the PAG contributed to the second meeting.

3. Results

Participant characteristics are outlined in Table 1. The participants were well educated, with nearly two-thirds having at least a Bachelors or equivalent first degree (65.6%). Approximately half were women (46.9%) and exactly half had children aged under 18 years old. Despite

were subsidiary to the research questions.

"The key message I got is that we have a system in place to kind of quantify quality of life for adults, but actually there's not one in place for children. And then, additionally from that, it's kind of how do we quantify it, like who do we ask? Do we ask children, do we ask adults? What sort of, you know, children do we ask them to picture?" (Participant 2 [P2], Focus Group [FG5]).

"I think it's about, again, like weighing up whether we should put more effects on quality of children and adolescents care over, say the length of life in general." (P4, FG2).

This subsidiary focus and misunderstandings necessitated further clarification by the facilitator prior to the discussion. Following this, participants showed a better understanding of the key research questions being discussed. Nevertheless, during the discussion, participants sometimes lost focus (e.g. by focusing on healthcare decision-making in general, rather than valuing health) and had to be brought 'back on-track' by the facilitator.

3.1.2. Understanding of health-related quality of life

While primarily designed as a warm-up exercise to discussing who should be asked to value child HRQoL, participants showed a sophisticated understanding of HRQoL. This was discussed in terms of what mattered to participants themselves, what they thought mattered to younger people, and how the importance of aspects of HRQoL may change over time. Participants showed an awareness that different people (including adults and young people) find different aspects of HRQoL important and some awareness of the 'disability paradox', that a child (or person) with health problems may report a greater level of HRQoL than adult or parents' proxy ratings (Albrecht and Devlieger, 1999).

"I think there might be times in, like, similar situations that where, like, the adult, and the parent might think that the child has a diminished quality of life but the child might not see it that way ... we would see it as affecting quality of life and the child might think that they're living like a full and, you know, great quality of life." (P2, FG5).

3.2. Who should Be asked?

3.2.1. Support for asking young people

The vast majority of participants supported asking or involving young people in some form and this typically differed by the age of the young person being considered. Four primary arguments were discussed relative to this position.

"I think children should be involved" (P6, FG2).

"... in terms of, like, actually giving the answer, I'd say that it's mostly for the child." (P2, FG5).

- (1) **Asking Children about Death.** Mixed views were observed on whether children should be involved in valuation tasks that involve trading off life years and thinking about death. Overall, there were more participants arguing against (n = 5) than for (n = 2) asking children about death. While one participant gave an ethical argument for not asking children about death, four others noted conceptual difficulties; that children do not understand the concept of death as well as adults.

"... it's not just an ethics issue, just think children don't really have that much experience of death or concepts of time so I feel like they might not necessarily understand what that means." (P7, FG3).

Arguments for involving children in valuation tasks involving death focused on its hypothetical nature and if done in a sensitive way it could

be acceptable.

"I think ethically it's personally fine to ask a child a hypothetical question. I mean, I don't see how that can really be morally bad. Hypotheticals." (P7, FG4).

- (2) **Young People Have a Right to Have a Say.** One of the most endorsed arguments (n = 9) for asking children was that young people have a right to have a say in decisions that affect them. No participants argued against this position. Participants felt fundamentally that children should have a say on issues that affect them at any age (beyond infancy), but some argued that greater weight should be afforded to the views of older versus younger children.

"I think children should still have that input and say it's their bodies and whatever health issues that they're facing whether it's mental or physical, I think that they should still be able to have a say, especially like older teenagers. You know, until they're in a position to pay taxes and contribute to, you know, the Government funding, they should still have a say." (P3, FG1).

- (3) **Adults Find It Hard to Imagine.** A few participants (n = 4) endorsed the argument that children should be asked because adults no longer know what it is like to be a child and would find it difficult to imagine this conceptually. One participant displayed a tension between this idea and adults having greater life experience.

"So for like, as a child, I personally think it would be best for them to give their opinion cos there's only them that knows how they feel and you don't know how it feels until you've actually experienced it and been, been through it. So, they would put like a value on what the, how they feel and what they would live with and what they wouldn't." (P1, FG1).

- (4) **Children Are Held Accountable in Other Circumstances.** One participant raised the argument that children are held legally accountable in other circumstances (citing the age of legal responsibility) and made the point that, by analogy, if we can hold them accountable for their actions in that context, why can't we involve them in valuation tasks?

"I think the, the age of legal responsibility is ten. So, if you use that argument, then, that if a ten-year-old murdered someone you can hold them accountable, then why can't they make their own decisions as well? It's a bit of a flip flop." (P7, FG4).

3.2.2. Support for asking adults

Most of the support for asking adults came in the context of asking them alongside young people, but there was a minority of participants who supported the idea of asking adults exclusively. Five arguments were discussed relative to support for asking adults.

"I think, you know, up to 16, it should be the adult's decisions. That's my personal experience to this as, you know, we've had it already" (P2, FG4).

- (1) **Taxpayer Argument.** The taxpayer argument was presented as one often cited as a justification for asking adult members of the public to conduct valuation tasks. In the two focus groups where this was discussed, no participants were supportive of this argument. More people (n = 4) were critical of this argument and spoke instead of the need for children to be included, despite their

reduced tax contribution. A few people's comments ($n = 3$) were coded as neutral and instead referred to a sense of confusion over why taxes were being brought into the argument at all.

"I mean, to be honest with you, it's quite a laughable argument, of course children don't pay tax, but it doesn't mean they're less important. You know, they're never going to pay tax because they're children, but they still feel pain and they still have a voice." (P5, FG7).

(2) Adults Make Important Decisions on Behalf of Children. One of the more endorsed arguments ($n = 7$) made for asking adults was that adults already make important decisions for children, have legal responsibility, and advocate on their behalf (especially parent/guardians). This was particularly the case when discussed in the context of the values informing funding decisions, which was seen as an important task that some participants believed may be better suited to adults. This argument was more salient for younger children.

"But again, I'm sure that a parent, my view is that typically a parent should always be consulted, up to that age point which we've not yet discussed." (P1, FG2).

(3) Task Too Complicated for Young People. Five participants voiced the opinion that the task of deciding which aspects of HRQoL they value over others may be too complicated for children, particularly those of a younger age. This was particularly the case when discussed in the context of a standard time trade-off (TTO) or discrete choice experiment (DCE) methodology.

"I don't think they would understand. If you said, right, OK, eight years of a good life, or ten years, you might not be able to walk, but you'd get an extra two years if ... I don't think they'd be able to grasp that concept." (P5, FG5).

(4) Children May Not Know What Is Best for Them. Six participants referred to the fact that children, and especially younger children, may not know what is best for them and their HRQoL over the long-term, when compared to adults. One participant suggested that children may be more susceptible to peer influence and another described how young people are still developing neurobiologically and this may affect their decision-making.

"One of the aspects we've not really covered is if I saw that my daughter couldn't walk at all that'd be awful, but she might not have the same understanding of how that would affect her and her quality of life ... if you're funding this you're looking at long-term difficulties and effects and if you can't walk that, that impacts children for the rest of their lives which they don't understand that concept I guess at a young ages, essentially." (P2, FG1).

(5) Life Experience. The most discussed argument related to asking adults was their greater degree of 'life experience'. Participants mostly used this as an argument to support asking adults ($n = 7$), but a couple also framed it as an argument against asking adults ($n = 2$), as increased experience could lead to bias.

"They're going to see it from a totally different perspective, I don't think that they fully appreciate. I'm not a, I can't, I'm not trying to be dismissive, and it would vary from child to child and age to age, but you know until you've lived a certain amount of life, I don't think you can fully appreciate the importance of it." (P1, FG3).

3.2.3. Support for asking adults and young people

There was broad support, articulated by approximately half of participants, for asking adults and young people combined to value children's health. Participants held different views about how much weight (or importance) should be placed on the contributions of adults versus younger people in this scenario, which differed depending on the age of the latter (with children given more weight as they get older). Further discussions were had on whether the adults and young people should be asked separately or dyadically, with arguments given on either side, including, for example, that parents of children could help them understand and complete the task. Two primary arguments were discussed in relation to the benefits of asking adults and young people.

"I'd probably say under ten, maybe weighted slightly more towards the adults and over ten, slightly the other way. So maybe sixty/fifty to the adults and sixty/fifty to the kids." (P3, FG5).

(1) Multiple Viewpoints Are Better. One argument used to support asking adults and young people ($n = 5$) was that multiple viewpoints are better than a single viewpoint. Occasionally, this included the suggestion of incorporating multiple viewpoints of people from different backgrounds (i.e., not solely differences in age), such as medical professionals and parents.

"So I think it needs to be a combination of things, whether that's medical professionals, parents and guardians, the person themselves who it is going to impact. But, it's, it's a hard one because I don't think any one person can make the best decision." (P4, FG2).

(2) Adults and Children Can Both Input into Decisions. A second argument endorsed ($n = 11$) for involving both adults and young people in valuation tasks was that they could both directly input into the choices and this would enhance the result. This was discussed in the context of adults (e.g., parents) helping, particularly younger, children with valuation tasks (e.g., ensuring understanding and reasoned responses), as well as support for adults listening to and considering a young person's view, while ultimately making the overall 'decision'.

"there still needs to be that input from the adult as well to like say explain the question more clearly if they don't understand and they might respond better having the parent and adult present as well." (P1, FG1).

3.2.4. Role of age, capacity, and maturity

Virtually all discussions about who to ask were contextualised based on the age of the children being considered and/or levels of cognitive capacity and maturity. Participants typically stated they would be more likely to involve children at older rather than younger ages. The rough age at which people said they would be happy to ask children about this topic varied, from as young as 3 years old (with assistance) to 16 years old. Participants were typically willing to ask younger children if this was accompanied by adult input (e.g., parental guidance). Other participants stated that age was too blunt a tool and it would rather depend on level of maturity or cognitive ability.

"But I think if you can get, establish an age where they're, like you say, emotionally mature, are able to empathise, cos a lot of these issues I personally haven't experienced in terms of like mobility, so that's me trying to put myself in someone else's shoes which is quite a difficult concept, so you need children to be old enough to be able to think that way. I had in my head before [P1 name] said about 10 as well, so that was quite, [P5 name] was saying 10 and [P1 name], but whether that's, that's right or not." (P3, FG1).

There was also discussion about the responsibility of researchers to

adapt methods to be delivered in an ‘age-appropriate way’, so that younger children’s views could be reliably and appropriately sourced and incorporated.

“I’ve actually got experience of doing research with children. And with the right methods and tools you can actually talk to them at a very young age, quite simple concepts and ways of doing things, but I think from the age of 3, 4, you can start measuring these sorts of things. They know what’s important to them at quite a basic level, and if you marry that up with some other questions for parents, I think it’s just as valid.” (P7, FG3).

3.2.5. Characteristics of people that should Be asked

Aside from a person’s age, participants discussed other desirable characteristics of people that they thought should be asked to value children’s health. These included people who had experience with children, with a lot of support for asking parents, and those with prior experience of ill health. Participants appeared to intuitively support the idea that people with experience would provide more ‘accurate’ or ‘enlightened’ responses. Participants also shared characteristics of people that they would not like to be asked in the context of valuing health in children. This included ‘the elderly’ (who could no longer advocate for children) and people with serious mental health problems or a diminished capacity to consent.

“... like maybe someone who was a parent, a carer or works closely with children, such as a teacher. Who knows the social side of children’s wellbeing as well. But, I think, maybe that’s just coming from having a child, I don’t know.” (P3, FG5).

3.3. Who should people think about (which perspective)?

3.3.1. Support for thinking about themselves

Most participants who discussed the issue supported the use of an ‘own’ perspective in valuation tasks, noting that you would get a ‘truer’ response and it reduces confusion for the respondent. Two main arguments were discussed in this regard.

“And you only ask people, based on themselves. Because then there’s no confusion about, are you asking about a ten-year-old, are you asking an adult, are you doing whatever. Just go back to basics.” (P4, FG4).

- (1) **Difficult to Imagine for Someone Else.** The most endorsed argument ($n = 7$) for using an ‘own’ perspective was that it was difficult to imagine for someone else (e.g., to imagine being a 10-year-old child). Participants expressed that it is easier to obtain a ‘factual’ response with an ‘own’ perspective and it avoids supposition. Only one participant argued against this argument, stating that adults could imagine because they have been younger before.

“I think you’re going to get a much more factual response if you ask the adult about their perceptions of what affects them, if they’re trying to put themselves into the mind of a 10-year-old child what you’ll get will be a product that is based perhaps, what their memories are of being a 10-year-old child, because you’ll have some imagination and you’ll have a lot of, make-believe is the wrong word, but supposition.” (P2, FG3).

- (2) **People Think About Themselves Anyway.** Two people raised an interesting supplementary argument for using an ‘own’ perspective, that even if people are asked to think about someone else in a valuation task, they think about themselves anyway.

“I think when you ask someone to put themselves in the shoes of somebody else, they just naturally think about their selves anyway, maybe at that

age. Or how, or how they understand someone at that age with how they were at that age. So, they’re essentially the same question, really, but just thinking about their past.” (P7, FG4).

3.3.2. Support for thinking about someone else (another child)

Amongst the minority support expressed for people thinking about someone else (another child) when valuing children’s health was two participants’ support for adults thinking about a 10-year-old child. One primary argument for thinking about another child was discussed.

“Ten’s a pretty good age if you’re asking an adult, I think ... I think you’d like to hope that most people would think kindly of a ten-year-old, and, and want to make the right choice for them, and be kind of benevolent to that fact.” (P6, FG4).

- (1) **It Is Children’s Health That Is Being Valued.** The one argument made by two people to support (adults) thinking about someone else (another child) was that it is children’s health being valued and responses may be different if you are thinking of yourself.

“Whereas, for the children if we did that say that cut-off and we’re, adults are answering for children they probably should be in the perspective of the child because they’re the ones that are gonna get the healthcare, or the benefits, or they’re the ones that in distress not you.” (P2, FG1).

3.3.3. Knowing it is Children’s Health That Is Being Valued

All but one of the participants who spoke on the issue of whether respondents should know it is children’s health (states) that are being valued was supportive of disclosure. This position was argued for on the basis of transparency and ethics (i.e., people have a right to know) and that responses may differ, so people should know what the values will be used for.

“I think if, if anything, if you’re being asked to make a, like, a choice that could impact somebody else, you should be given sort of full disclosure. Anyway, I don’t really feel comfortable with the idea [laughs] of, like, making decisions on someone when you don’t actually know enough about them. And like, like the last person said, if it, if it impacts a child’s life and, you’ve made that decision, surely you should have been able to have the knowledge it was a child that you’re impacting.” (P3, FG2).

4. Discussion

The current study used a focus group methodology to explore views of informed adult members of the UK public about two key normative issues in the valuation of child HRQoL: Who should be asked to provide the values and who should they think about (i.e., using what perspective)? In doing so, it adds to a growing evidence base on stakeholder perspectives’ on valuing HRQoL for children and young people, for example, as part of the ongoing EQ-5D-Y-3L valuation studies (Devlin et al., 2022a).

Key findings from this study are indicative of a lack of UK public support for some aspects of the recent EQ-5D-Y-3L valuation protocol (Ramos-Goni et al., 2020). For example, the current sample of adult UK taxpayers did not intuitively support the ‘taxpayer argument’ used to justify asking adults (who bear the primary tax burden funding public healthcare) to value health for children (Lipman et al., 2021b). Nor was there much support for the idea that adults should be asked to think about another person (i.e., a 10-year-old child) in valuation tasks, because of perceived problems with that approach. Instead, participants believed that young people should be involved in HRQoL valuation in some form, with their degree of involvement depending on their age and/or observed cognitive capacity or level of maturity. This approach was advocated based on a number of arguments, one of the most

endorsed being that young people should have a right to have a say in decisions that affect them. While there was a recognition that tasks may be too complicated for (younger) children, a number of participants voiced that the onus was on researchers to develop better methods to enable their involvement.

Regarding perspective, amongst those participants that the topic was discussed, the modal response was support for an 'own' perspective as a way of minimising abstraction and confusion. This opinion needs to be viewed in context, alongside participants views on who to ask, that is participants were generally supportive of young people being involved in the valuation task (as well as adults) and that when they were involved they would be imagining themselves living in the health state. Nevertheless, this finding is consistent with other qualitative work exploring perspective-taking in child HRQoL valuation, where an 'own' perspective was similarly advocated amongst an independent sample of the UK public (Powell et al., 2021). This means that, while the decision was made amongst health economists and allied experts to use a 10-year-old child perspective in the valuation of the EQ-5D-Y-3L, it may not be supported by the participants who are being recruited to complete those tasks.

While most participants supported the idea of asking young people, there was not universal support for exclusively asking young people. Instead, some participants argued for the involvement of adults due to their greater 'life experience' and responsibility for making important decisions. In fact, across-the-board, when speaking on the issue there was a tendency for participants to support the idea of asking a combination of young people and adults. The PAG were also supportive of this approach, whilst also noting the practical issues that may need to be solved. Participants argued for involving adults and young people based on the benefits of incorporating multiple perspectives and that having an adult or young person complete the task could ameliorate any deficiencies in choosing one sample alone (e.g., adults could help younger people respond in an informed way; children could help adults better understand what their views are). Similarly, across most participants, greater weight was afforded to the contribution of older children and adolescents than younger children. These views are consistent with recent US HTA stakeholder work as part of the valuation of the EQ-5D-Y-3L which recommended the incorporation of adolescent values (Nazari et al., 2022). Taken together, they suggest that initiatives to involve older adolescents as part of a mixed sample in the valuation of HRQoL for children, using an 'own' perspective, may be a progressive step in the right direction, according to views of stakeholders and the UK public (Rowen et al., 2022). A related point is the extent to which separate versus combined value sets are warranted as a function of age and this is a question for further consideration and research (e.g., with policymakers).

Alternatively, the idea of dyadic interviews (e.g., involving parents) was raised as one that had potential, but is one that – to the authors' knowledge – has only been explored in the context of acquiring HRQoL data from children, rather than health state valuation (Ungar, 2011; Ungar et al., 2006, 2012). This is presumably due to a combination of concerns around the involvement of children and young people in valuation tasks and/or the perceived respondent bias that may occur on behalf of a young person if an adult (e.g., their parent) is taking part with them. However, as well as its limitations, dyadic interviewing has potential advantages that should not be discounted, such as the ability for a parent to act as an 'enabler', providing the child with clarifications, past examples, and positive feedback, that may enhance data collection (Ungar et al., 2006). A dyadic approach to health state valuation was recently experimentally trialled in peer-to-peer student dyads, with little impact on decision quality (Attema et al., 2020). A further, and potentially complementary, research direction is to explore ways in which health economics valuation methods could be adapted to be more inclusive and 'child friendly'.

Finally, there was broad support in this sample for the disclosure of information to participants as part of health state valuation tasks, which

are then used to score the health of children and young people. In particular, informing them that the health states being described are for children and, potentially, what their responses would be used for (i.e., to generate utilities to inform outcomes in cost-effectiveness analyses). The PAG were also very supportive of this stance, supporting an approach of full transparency, and noting an ethical justification for fully informing participants. While prior research has shown mixed views and counterarguments exist, such as the potential emotional investment of knowing it is about children (Powell et al., 2021). This represents an important area for discussion and consideration moving forward in this field.

4.1. Limitations

The present work is the first time members of the UK public have been consulted about their views on who should be asked to value HRQoL for children and adolescents. Best practice has been followed in conducting the work, including archiving and following an a priori protocol, following recognised qualitative methods, using methods of quality assurance, and involving patient and public collaborators in the study. Nevertheless, as with all studies, several limitations are apparent.

First, while the researcher was cognizant of and took steps to avoid influencing participants by presenting information and arguments in an unbiased manner and not endorsing any particular stance, we cannot discount that the way information was presented or the way topics were discussed may have influenced responses (including what was not said). This is true of all qualitative research and methods of assuring quality were put in place to help mitigate this. Further, arguments for and against different approaches were communicated in relative lay terms to participants and thus we cannot claim that they fully understood these arguments at the level of economic theory.

Second, this study recruited only adult participants to talk about an issue that affects young people, without inclusion of young people. Furthermore, all participants were recruited from England and so the findings should be interpreted within this sociocultural context. This is a clear limitation of the work and is defensible only in so much as this was an initial qualitative study into this topic to develop insights into the views of the UK adult public. Complementary research on this topic should seek to incorporate the views of young people and those in other countries and/or cultures (Åström et al., 2022).

Third, the focus groups for this study were conducted online, rather than face-to-face, and it is possible this may have had an impact on the data acquired. While there are clear advantages to online qualitative methods, such as recruiting from a more diverse geographical area, there are also disadvantages, such as reduced rapport and potential inclusion.

Finally, within the focus groups and as evident from the thematic coding, there was more discussion devoted to who to ask than who to think about (i.e., which perspective). This occurred for a number of reasons. First, the latter was judged as conceptually more difficult than the former and it is necessary to fully understand the former before discussing the latter. Accordingly, the topic guide was designed to discuss 'who' first and this often left less time in the focus groups for discussing 'which perspective'. Second, prior qualitative research by this research group has explored UK adult public's views on perspective when valuing child health (Powell et al., 2021). Therefore, their views on the question of 'who' is more novel and was given prominence in this study.

5. Conclusions

To the authors' knowledge, this is the first comprehensive qualitative study to ask members of the UK adult public for their perspectives on who should be asked to value HRQoL for children and with what perspective. As part of the process, members of the public were 'informed' about health state valuation and key normative issues in

valuing HRQoL for children, using bespoke informational resources and qualitative clarifications.

Key findings from this study suggest that participants think young people should be involved in valuing their HRQoL in some form, but the precise age at which this should occur is difficult to determine. There was broad support for a dyadic approach, involving young people and adults in health state valuation, due to the perceived benefits of inclusion and multiple perspectives. Very few participants supported asking adults exclusively. The most popularly endorsed argument for asking children was that they have a right to have a say in things that affect them. Arguments endorsed by participants for asking adults included that they had greater 'life experience' and already make important decisions for children. Most participants, who spoke on the issue, thought that asking children about death was problematic, while participants were critical of the 'taxpayer argument' for asking adults.

In the context of involving young people in health state valuation, the majority of participants voiced support for an 'own' perspective over-and-above thinking about another, such as a child, due to the reduced abstraction and confusion the latter could cause. Further, participants and the PAG were supportive of full disclosure in informing participants that they were valuing child health states and what the responses may be used for.

Taken together, these results suggest that members of the UK public sampled in this study did not intuitively support the normative positions adopted in the recent EQ-5D-Y-3L valuation protocol, (Ramos-Goñi et al., 2020) which includes exclusively asking adults, taking the perspective of a younger child, and justifying the former with a 'taxpayer argument'. In contrast, the results suggest including adults and children in valuation, with the proviso that the children are of an appropriate age and level of maturity, and that an own perspective is used wherever possible.

CRedit authorship contribution statement

Philip A. Powell: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Resources, Visualization, Writing - original draft, Writing - review & editing. **Donna Rowen:** Conceptualization, Formal analysis, Funding acquisition, Investigation, Writing - review & editing. **Anju Keetharuth:** Formal analysis, Funding acquisition, Investigation, Writing - review & editing. **Clara Mukuria:** Formal analysis, Funding acquisition, Writing - review & editing.

Data availability

The data that has been used is confidential.

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Appendix A. Supplementary data

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