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Powell, P.A. orcid.org/0000-0003-1169-3431, Rowen, D. orcid.org/0000-0003-3018-5109, Keetharuth, A. orcid.org/0000-0001-8889-6806 et al. (2 more authors) (2024) Who should value children’s health and how? An international Delphi study. Social Science & Medicine, 355. 117127. ISSN 0277-9536

https://doi.org/10.1016/j.socscimed.2024.117127

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Social Science & Medicine 355 (2024) 117127

Contents lists available at ScienceDirect

Social Science & Medicine

journal homepage: www.elsevier.com/locate/socscimed

Who should value children's health and how? An international Delphi study

Philip A. Powell [a,*], Donna Rowen [a], Anju Keetharuth [a], Clara Mukuria [a], Koonal Shah [b]

[a] Sheffield Centre for Health and Related Research, University of Sheffield, UK
[b] National Institute for Health and Care Excellence, London, UK

ARTICLE INFO

Handling editor: Joanna Coast

ABSTRACT

Valuing child health necessitates normative methodological decisions on whose preferences should be elicited and who should be imagined as experiencing impaired health. Formal guidance is limited and expert consensus unclear. This study sought to establish the degree of consensus among expert stakeholders on normative issues of who to ask and who should be imagined when valuing child health (7–17 years) to inform UK health technology assessment. Sixty-two experts (n = 47 in Round 2) from 18 countries participated in a modified, two-round online Delphi survey (Round 1: May-June 2023; Round 2: September-October 2023). Participants were expert stakeholders in child health valuation, including academics (n = 38); industry/consultancy representatives (including the charity/not-for-profit sector; n = 13); and UK policy/government representatives (n = 11). The Delphi survey was modified between rounds and consisted of 9-point Likert, categorical, multiple-choice, and free-text questions on normative issues in valuing child health. Responses were analysed descriptively and thematically. An *a priori* criterion of ≥75% agreement was established for formal consensus, while areas approaching consensus (≥70% agreement) and without consensus were identified as a future research primer. Consensus was observed that older adolescents (aged 16–17 years) and adults (18+ years) should be asked to value child health states. There was consensus that the former should think about themselves when valuing the health states and the latter should imagine a child of some form (e.g., imagining themselves as a child or another hypothetical child). However, no consensus was evident on what form this should take. Several other methodological issues also reached consensus. These findings are largely consistent with recent views elicited qualitatively from members of the public and other stakeholders on normative issues in valuing child health. The results mean that, contrary to what has been done in previous child health valuation studies, efforts should be made to involve both older adolescents (16+ years) and adults in child health valuation.

1. Introduction

Valuing health states is a core facilitative ingredient for health technology assessment (HTA), and enables the generation of quality-adjusted life years (QALYs) for cost-effectiveness analyses (Brazier et al., 2016). Health state valuation involves quantitatively eliciting people's relative preferences for living in different states of health. For HTA, this information is typically elicited using trade-offs with length of life (or survival), which allows for a relative estimation of QALYs for different health states (using the same denominator), anchored at 0 (dead) and 1 (full health), with values lower than 0 defined as states appraised as worse than dead (Brazier et al., 2016).

In the UK HTA process, and as recommended by the National Institute for Health and Care Excellence (NICE) (National Institute for Health and Care Excellence (NICE), 2022), health state values for adult health states are elicited using an established choice-based method, with a sample of the adult general public, representative on key demographic criteria (e.g., age, sex, ethnicity, socioeconomic status) (Rowen et al., 2023). These participants are asked to complete the valuation tasks thinking about themselves in impaired health. Responses are then averaged, and if elicited for a preference-weighted measure are modelled to produce a 'societal' value set for the measure. However, similar methodological guidance on valuing child and adolescent health states is not provided, with the NICE HTA methods guide stating that:

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[*] 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).

https://doi.org/10.1016/j.socscimed.2024.117127
Received 15 March 2024; Received in revised form 13 June 2024; Accepted 9 July 2024
Available online 11 July 2024
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"there are methodological challenges when developing value sets for children and young people" (National Institute for Health and Care Excellence (NICE), 2022).

Amongst the choices that need to be made when valuing health of children and adolescents are two normative questions (Rowen et al., 2020). First, who should be asked to complete the valuation task? Adults could be asked to value child and adolescent health states, children and adolescents (of a specified age) could be asked to do so, or a combination of both adults and children could be asked (Rowen et al., 2022). Second, who should be imagined as experiencing impaired health (also referred to here as 'which perspective')? Participants could be asked to value child and adolescent health states thinking about themselves experiencing impaired health (i.e., an 'own' perspective), or thinking about another person in those health states, such as a younger child (i.e., an 'other child' perspective) (Powell et al., 2021). Alternative permutations are possible and further intricacies are evident, such as at what age children should be asked to complete valuation tasks or what aged child should be thought about when completing the tasks. These decisions matter, as different choices elicit different values and thus different QALY estimates for use in HTA (Lipman et al., 2021a; Prevolnik Rupel et al., 2021). Therefore, guidance on these methodological issues would be helpful to standardise how health state values are elicited for children and adolescents.

One notable attempt to standardise the process of valuing child and adolescent health is the valuation protocol for the EQ-5D-Y-3L, published in 2020 (Ramos-Goñi et al., 2020). For valuing EQ-5D-Y-3L health states (which has a target age range of 8–15 years (Devlin et al., 2022)), the protocol recommended the use of adult general public preferences thinking about a 10-year-old child. While no rationale was provided for the focus on a 10-year-old child, a 'taxpayer perspective' was cited in support of asking adults (i.e., as the adult public shoulder the predominant tax burden for funding the health service their preferences should be prioritised (Ramos-Goñi et al., 2020)). A range of country-specific value sets for the EQ-5D-Y-3L have been produced using this protocol, with more ongoing (Shiroiwa et al., 2021; Kreimeier et al., 2022; Prevolnik Rupel and Ogorevc, 2021; Roudijk et al., 2022; Yang et al., 2022; Rencz et al., 2022; Fitriana et al., 2022; Dewilde et al., 2022a). However, several papers are now questioning the basis of the normative decisions in the EQ-5D-Y-3L protocol (Lipman et al., 2021b; Nazari et al., 2022; Powell et al., 2023; Xie et al., 2024).

As these questions are recognised to be normative (i.e., value-based judgements (Devlin et al., 2022)), while they may be informed by research (e.g., on children's cognitive ability), there is no objectively correct answer and decisions are made through normative consensus. An understanding of consensus in this area is currently lacking and input from key stakeholders in the valuation process is likely to be of value, including academic experts, HTA policy representatives, and members of the public themselves (Devlin et al., 2022). Establishing areas of consensus will help to ensure that normative decisions made in the valuation of child and adolescent health appease the majority of stakeholders involved in the process. Establishing areas of lack of consensus will identify where additional information gathering, work, and debate is needed. A recent qualitative study elicited informed views of the UK public on the normative questions of who to ask and who to imagine in child and adolescent health valuation (Powell et al., 2023). However, no study to date has established quantitatively the degree of consensus on these normative issues in a substantial sample of professional stakeholders.

The present study was designed as the first of its kind to systematically establish where there is (and is not) normative consensus on the issues of who to ask and who to imagine in valuing child and adolescent health. A modified online Delphi procedure – a well-established consensus-based methodology (Murphy et al., 1998) – was used to address this research aim. Academics, industry experts, and policymakers were invited to participate. While international in scope, the context of the exercise was valuing child and adolescent health states (aged 7–17 years) for HTA in the UK. No *a priori* predictions were made about where consensus (or lack of) would be observed, but it was hoped that insights from this Delphi exercise would be of use to international researchers and UK policymakers in moving towards recommendations of normative positions in the valuation of child health.

2. Methods

2.1. Design

A protocol was archived online prior to data collection (doi.org/10.15131/shef.data.20424915.v1). A modified Delphi methodology was adopted to "explore or expose underlying assumptions or information leading to different judgements" (Hasson et al., 2000). The Delphi method is recommended in areas without a clear and *prima facie* consensus (Murphy et al., 1998). The research was conducted and managed online on Qualtrics (an online survey platform), with a panel of stakeholders in HTA. The Delphi was designed to feature up to three iterative rounds. However, the research team, in consultation with advisors from the funding body, made the decision to restrict the survey to two rounds based on perceived diminishing returns relative to additional resource cost and a low likelihood of achieving consensus on core outstanding issues (Hasson et al., 2000). Modifications were made to the 'classical' Delphi design (Hasson et al., 2000), including the use of pre-formed quantitative questions in Round 1; presenting results to participants qualitatively as well as quantitatively; and the addition of questions between rounds based on feedback from participants and consultation within the research team. The research was designed and reported in concordance with the Recommendations for the Conducting and Reporting of Delphi Studies (CREDES) (Jünger et al., 2017).

2.2. Participants and recruitment

Target participants for the Delphi panel consisted of professionals with knowledge of and/or applied expertise in valuing child health. These included international health economists and allied researchers, industry/consultancy representatives (including charities/not-for-profit organisations), and UK policymaker representatives from NICE and the UK Department of Health and Social Care (DHSC). A list of 102 experts was drawn-up by the research team via targeted literature searching; advice from NICE, DHSC, and the National Institute for Health Research (NIHR) Policy Research Unit in Economic Methods of Evaluation in Health and Social Care Interventions (EEPRU) advisors; and screening the member lists of key stakeholder groups (e.g., the EuroQol Research Foundation). Based on estimated attrition, this target sample was deemed sufficient to exceed the modal number of participants in the final round of a Delphi (11–25 people (Diamond et al., 2014)). Potential participants were invited to participate in the by e-mail. Only those who took part in Round 1 of the Delphi were invited to participate in the subsequent round. Participants were not reimbursed for their time.

2.3. Delphi survey and procedure

A flowchart describing the study process is in Fig. 1. Informed consent was acquired at each round. The 72-question Round 1 Delphi survey (Supplementary File A) was designed to feature a range of questions eliciting expert views on issues relevant to the normative choices made in valuing child health, including their perceived endorsement of underlying arguments. The survey included Likert-type questions, using a 9-point scale (1–3 = do not agree; 4–6 = neither agree nor disagree; 7–9 = agree, (Niederberger and Spranger, 2020)), and qualitative free-text questions, allowing participants to provide reason(s) for their responses, further information and feedback, and identify any areas that were missing for future Delphi rounds. Participants were given 3-weeks to complete the Round 1 Delphi survey, with reminders at 1-week, 72-h, and 24-h before the deadline.

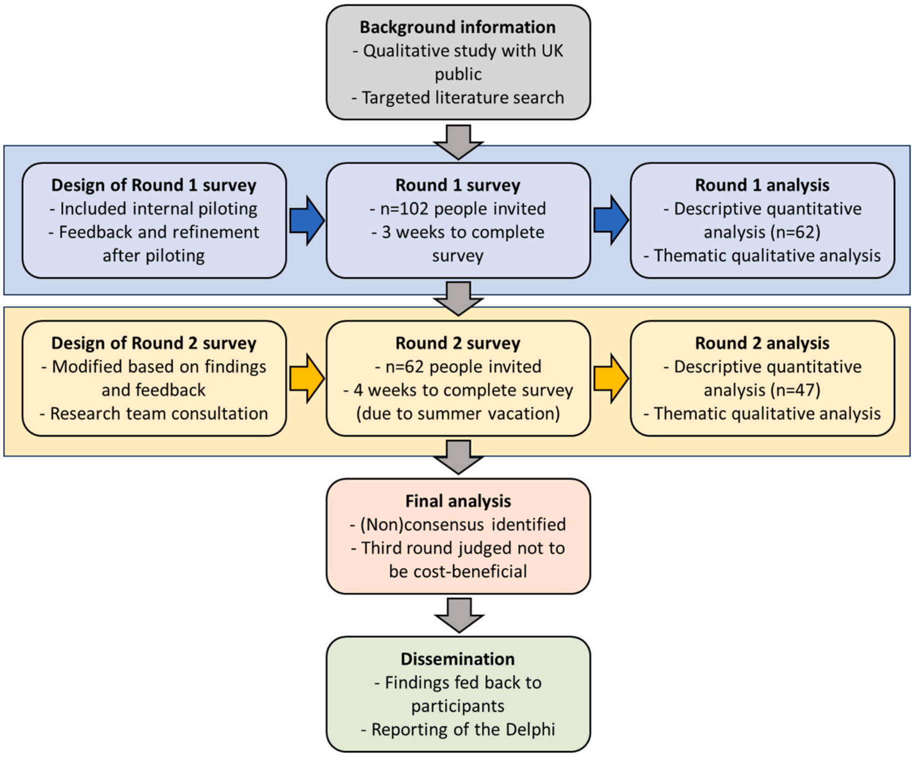

Fig. 1. Flowchart of the Delphi study.

To help contextualise their opinions, participants were provided with two sources of information prior to completing the survey: (i) the views of a sample of the UK public on who to ask and who to imagine in child health valuation, based on a recent qualitative study (Powell et al., 2023); and (ii) information on the existing arguments for and against different normative positions in child health valuation, based on a targeted literature search (Rowen et al., 2020, 2022; Powell et al., 2021; Prevolnik Rupel et al., 2021; Ramos-Goñi et al., 2020; Lipman et al., 2021b; Nazari et al., 2022; Reckers-Droog et al., 2022; Kwon et al., 2022; Mott et al., 2021, 2022; Kreimeier and Greiner, 2019; Bailey et al., 2022; Khadka et al., 2019; Petrou, 2022; Rogers et al., 2021; Åström et al., 2022; Dalziel et al., 2020; Dewilde et al., 2022b; Crump et al., 2018; Devlin, 2022) (this included a label and extended description for each argument). Additionally, as part of the background information, participants had the option to view an informative and lay-friendly video on child health state valuation and the normative issues involved developed in a recent study (Powell et al., 2023), but this was not compulsory. Prior to the questions on normative issues in child health valuation, participants answered background questions on basic sociodemographics, profession, and self-rated expertise related to the valuation of health for children (see Supplementary File A). The Round 1 survey and background information was piloted internally by five members of University staff, unaffiliated with the current project, and revised prior to launch.

Responses from the Round 1 survey were used to inform the 48-question Round 2 survey (Supplementary File B), with areas of consensus omitted from further investigation and areas of non-consensus reassessed. Questions were modified and added based on participant responses and feedback, in consultation with the research team, to help clarify Round 1 responses. This included the addition of categorical questions, as well as Likert scales. Findings from the Round 1 survey were summarised and presented to participants quantitatively (i.e., graphically) and/or qualitatively (i.e., descriptively), alongside the summarised opinions of the UK public. While participants were initially given 3-weeks to complete the Round 2 survey, this was extended by an additional week, due to the summer vacation period. Participants were subsequently provided with a summary of the findings from Round 2 by email. Ethics approval for this study was obtained from the host institution (reference number: 046269).

2.4. *Analysis*

Quantitative responses were descriptively analysed based on percentage (dis)agreement and measures of central tendency (mean, median, and – where relevant – mode). An *a priori* criterion for consensus of $\geq$75% of participants either endorsing an option (categorical items) or agreeing or disagreeing with a statement (Likert items) was established (Diamond et al., 2014). The percentage of $\geq$75% was chosen as the median choice for establishing consensus in a systematic review of Delphi studies (Diamond et al., 2014). Any consensus responses in the range of $\geq$70% and $<$75% were described as “approaching consensus”, as a 70% cut-off is regularly used in other Delphi studies and represents agreement of over two-thirds of respondents (Audag et al., 2023; Shearsmith et al., 2020; Campbell et al., 2018). Quantitative data was descriptively analysed using Excel and R v4.2.2. Qualitative responses were qualitatively synthesised thematically and narratively summarised (Dimairo et al., 2018). Qualitative analysis was conducted by the primary researcher, with thematic coding checked for sense and agreement by another researcher.

3. Results

Sixty-two participants (60.8% response rate) took part in Round 1 of the Delphi survey and 47 participants (75.8% response rate) participated in Round 2. One partial response was recorded in Round 1, otherwise all data were complete. Participant characteristics are provided in Table 1. Responses were received from 18 countries (with concentrations in the UK, Australia, Netherlands, and United States). The Round 1 sample included 38 academics, 13 people working in industry, consultancy, or not-for-profit organisations, and 11 people working in policy or government. Overall, the sample had a high degree of experience, with a mean of 16.63 (min = 0, max = 37) years working

Table 1
Participant characteristics.

	M (SD) |N (%)	
	Round 1 (n = 62)	Round 2 (n = 47)
Age (years)[a]	46.66 (11.04)	45.95 (11.07)
Gender		
Woman	29 (46.8)	19 (40.4)
Man	33 (53.2)	28 (59.6)
Country		
United Kingdom (UK)	21 (33.9)	14 (29.8)
Australia	7 (11.3)	6 (12.8)
Netherlands	7 (11.3)	6 (12.8)
United States (US)	7 (11.3)	4 (8.5)
Canada	2 (3.2)	2 (4.3)
Germany	2 (3.2)	2 (4.3)
Indonesia	2 (3.2)	2 (4.3)
Slovenia	2 (3.2)	0 (0.0)
South Africa	2 (3.2)	2 (4.3)
Spain	2 (3.2)	2 (4.3)
Other (see legend)[b]	8 (12.9)	7 (14.9)
Primary profession		
Academic	38 (61.3)	31 (66.0)
Industry/consultancy/not-for-profit organisations	13 (21.0)	10 (21.3)
Policy/government (UK only)	11 (17.7)	6 (12.8)
Years' experience in health economics	16.63 (8.54)	15.94 (8.34)
Self-declared experience		
I have experience/knowledge in health economics, but no experience/knowledge of health state valuation in adults or children	1 (1.6)	0 (0.0)
I have experience/knowledge in health economics and health state valuation in adults, but no experience/knowledge of health state valuation in children	5 (8.1)	3 (6.4)
I have experience/knowledge in health economics and a knowledge of health state valuation in children, but no applied research or practical experience with the latter	15 (24.2)	11 (23.4)
I have experience/knowledge in health economics and applied research or practical experience of health state valuation in children	41 (66.1)	33 (70.2)

[a] N = 56 in Round 1 (6 people preferred not to disclosure their age); N = 43 in Round 2.

[b] The following additional countries had one participant in Round 1: China, France, Hungary, New Zealand, Norway, Poland, Singapore, Sweden (the participant from New Zealand did not participate in Round 2).

in health economics. All participants had experience or knowledge of health economics. Fifty-six (90.3%) participants self-reported a knowledge and/or practical experience of health valuation in children. Of those who did not, four were UK policymakers who are likely to be involved in decision-making in this area and two were academics with a known affiliation to projects on child health measurement and/or valuation. Five of the six had knowledge/experience of health valuation per se and the other was a UK policymaker. Thus, it was decided to retain them in the primary sample. Differences in findings with these six participants removed are outlined in Supplementary File D. The median duration of survey completers was 83.3 min in Round 1 and 54.4 min in Round 2.

A complete tabulation summarising all responses to questions asked across the two Delphi survey rounds is included in Supplementary File C. Results presented here relate to the whole sample, but differences at the consensus thresholds for UK-only participants and policy-makers only are provided in Supplementary File E and Supplementary File F. An overview of issues reaching consensus (≥75% of responses), approaching consensus (≥70%, <75%), and with no consensus is summarised in Table 2.

3.1. Who should be asked?

3.1.1. Round 1

Consensus was achieved on asking adults (18+ years) and older adolescents (16–17 years) to value child health states, both when trading off life years and not trading off life years (Fig. 2). No consensus was evident in asking children younger than 16 years. While a notable number of participants (n = 12) chose not to answer the questions on minimum age for inclusion in the valuation sample, those who did reported a median of 13.5 years (joint mode = 12 and 16 years) when trading off life years and 10.0 years (mode = 10 years) when not trading off life years.

No consensus was achieved on questions on sampling, nor on arguments for or against asking children to value health states. Regarding arguments **for** asking children, *empowerment/right to have say* approached consensual endorsement (70.5%) and the least endorsed argument was *adults will not be directly impacted* (21.7%). Regarding arguments **against** asking children, the strongest argument endorsed was *cognitive ability, understanding, and/or complexity of the tasks* (65.0%). The least endorsed argument was the *taxpayer perspective* (21.7%).

In the qualitative comments, the primary reason participants provided for their answer to who should be asked was the complexity of the tasks and young people's requisite level of cognitive ability, experience, and/or maturity to reliably complete them. Participants made the distinction between valuation methods that did and did not involve trading off life years, trending towards support for a younger minimum age for the former, due to both ethical concerns and conceptual understanding. Mixed views were given on sampling for parental status, with most comments preferring a representative sample of the general population that would include parents. Mixed views were also obtained for recruiting based on experience. Some confusion was observed over the question on a combined sample (with a single value set) versus separate value sets representing the preferences elicited from children and adults, prompting a need for clarification.

Qualitative views shared on the arguments for and against asking children to value health were sparse and mixed. Eight participants disagreed with the premise of *adults do not understand or experience children's health states*, noting that all adults have been children at some point. Ten participants disagreed with the argument *adults will not be directly impacted* as healthcare resource allocation budgets are not ring-fenced for children and adults could be impacted as caregivers. An additional argument was suggested that it *may not be in children's best interests to value the health states*. Amongst the other comments, one participant noted that we had not explored dyadic valuation approaches in the Delphi survey.

3.1.2. Round 2

Several modifications were made for the Round 2 survey on who to ask (Supplementary File B). First, due to potential heterogeneity, the age groups for younger children (7–15 years) were disaggregated and questions were asked about including children at each age. Clarifications were made to other methodological questions (e.g., about sampling) to help with understanding and these were made categorical questions (rather than 9-point agreement scales). A question was added on dyadic valuation (i.e., what minimum age could a child be included in valuation with their parent/guardian present?). Arguments were refined based on feedback, for example *adults will not be directly impacted* was revised to *children will be more impacted by the results than adults* and an additional argument was added: *it is not in children's best interests to value child health states*. Finally, in order to help facilitate consensus, rather than endorsement on a 9-point scale, participants were asked to select up to 3 arguments they thought were the most important or persuasive.

Consensus was achieved that children aged 10 years and younger should not be included in child health valuation tasks that involve

Table 2
Summarising areas of consensus in the Delphi.

	Consensus (+) | Approaching consensus (/) | No consensus (−)	+	/	−
Who should be asked?				
Age	Older adolescents (aged 16-17 years) and adults (aged 18+ years) should be included when valuing child health states.	✓		
	Adolescents aged 15 years old should be included if **not** trading off life years.	✓		
	Children aged ≤ 10 years should **not** be included if trading off life years.	✓		
	Children aged 11 years should **not** be included if trading off life years.		✓	
	Children aged 12 to 15 years should be included if trading off life years.			✓
	Children aged 7 to 14 years should be included if **not** trading off life years.			✓
Other methods questions	If adults are valuing the child health states, recruit a general population sample and not exclusively those with direct experience with children.†	✓		
	Involving children (of a specified minimum age) in dyadic child health valuation (i.e., with their parents).			✓
	How to analyse and produce value sets when adults and older adolescents are both recruited in the same child health valuation study.			✓
Arguments	'Children will be more impacted by the results than adults' and 'adolescent preferences can be combined with adult preferences' are **less** important arguments **for** including children.	✓		
	'Taxpayer perspective', 'logistical challenges to recruiting children', 'consistency with asking adults to value adult health states', and 'it is not in children's best interests to value child health states' are **less** important arguments **against** including children.	✓		
	'Adults do not understand or directly experience children's health states' is a **less** important argument **for** including children.		✓	
	Empowerment/right to have a say' and 'children consider different things as important to adults' are important arguments **for** including children.		✓	
	Importance of other arguments for and against including children in child health state valuation, not otherwise mentioned.			✓
Who should be imagined?				
Perspective	Adults (18+ years) valuing child health states should adopt some form of 'child' perspective.†	✓		
	If adolescents (aged 10-17 years) are valuing child health states they should adopt an 'own' perspective.	✓		
	Exactly what perspective adults should take if valuing child health states (e.g., an 'own child' vs. 'other child' perspective).			✓
	What specific aged child adults or older adolescents should think about if adopting a child perspective.			✓
Other methods questions	Participants should know they are valuing child health states.	✓		
	Participants should be told that their responses may indirectly influence funding decisions in children's healthcare.		✓	
Arguments	'Participants have a right to know it is about children' is an important argument **against** the use of an 'own' perspective (for adults).	✓		
	'Veil of ignorance/bias' and 'adults are less willing to trade off life years for children than adults' are **less** important arguments **for** adults taking an 'own' perspective.	✓		
	'Difficult to make decisions for others' is a **less** important argument **for** adults and older adolescents taking an 'own' perspective.	✓		
	'Consistency with prior research in valuing child health' is a **less** important argument **against** adults and older adolescents taking an 'own' perspective.	✓		
	'Consistency with adults (18+ years) using an 'other child' perspective' is a **less** important argument **against** older adolescents taking an 'own' perspective.	✓		
	'The 'Other child' that is imagined can vary' and 'difficult to make decisions for others' are **less** important arguments **for** younger adolescents taking an 'own' perspective.		✓	
	'Description of health states more appropriate for a younger 'child' perspective' is a **less** important argument **against** older adolescents taking an 'own' perspective.		✓	
	Importance of other arguments for and against taking an 'own' perspective in child health state valuation, not otherwise mentioned.			✓

Consensus defined as ≥ 75% responses. Approaching consensus defined as ≥ 70% responses (but not reaching consensus). Unless otherwise specified, 'adults' = 18+ years old; 'older adolescents' = 16–17 years old; 'younger adolescents' = 10–15 years old. †Consensus threshold reached in those participants not explicitly against adults valuing child health states. For full wording of questions please refer to Supplementary File A and B.

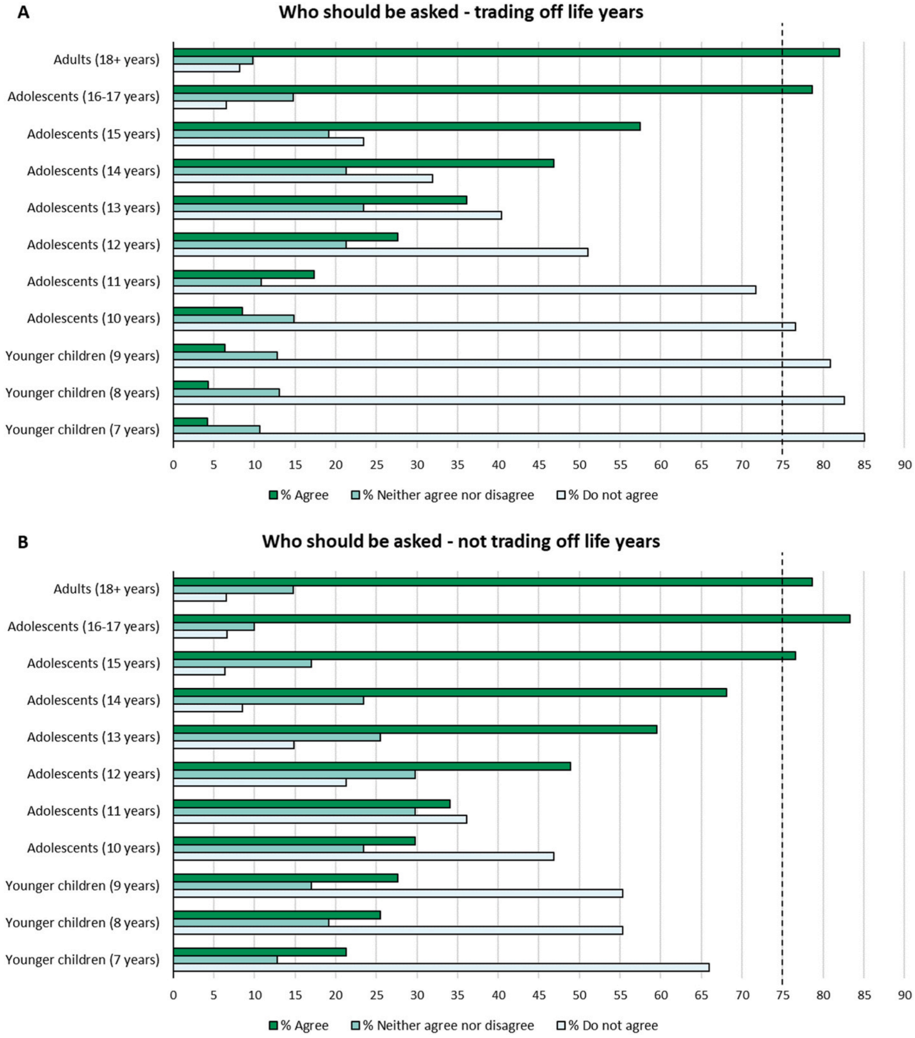

Fig. 2. Percentage agreement on who should be asked to value the child health states, involving a method that does (A) or does not (B) involve trading off life years. Note: Threshold for Delphi consensus set at ≥ 75%. Responses for adults (18+ years) and older adolescents (16–17 years) taken from Round 1 of the Delphi survey. Responses for all other ages taken from Round 2.

trading off life years (Fig. 2), and for 11-year-olds this approached consensus (71.7%). There was also consensus that 15-year-olds should be included in valuation if not trading off life years (76.6%). When recruiting adults to value child health states, there was an approaching consensus view (73.3%) that this should be a general population sample and not exclusively those with direct experience with children. This was a consensual view amongst those who were not against adults valuing child health states (80.5%).

Regarding arguments **for** asking children (Fig. 3), the arguments *children will be more impacted by the results than adults* (80.9%) and *adolescent preferences can be combined with adult preferences* (76.7%) showed consensus in not being selected as important. *Children consider different things as important to adults* (74.5%) and *empowerment/right to have a say* (72.3%) approached consensus as being selected as important.

Regarding arguments **against** asking children, *logistical challenges to recruiting children* (76.6%), *taxpayer perspective* (89.4%), *consistency with asking adults to value adult health states* (95.7%), and *it is not in children's best interests to value child health states* (95.7%), were consensually all not selected as important. While not reaching the consensus threshold, *cognitive ability, understanding, and/or complexity of the tasks* (68.1%) and *ethics/asking about death* (66.0%) were most commonly selected as important.

Qualitative comments made in Round 2 were broadly similar to that of Round 1, with primary considerations for including children in health state valuation being the complexity and feasibility of the task, relative to children's cognitive ability, and the ethics of trading off life years. Thirteen participants left a comment on dyadic valuation; their views were mixed. Four participants saw dyadic valuation as having great potential. Two participants wanted to know more about its feasibility through research, and seven others were sceptical, citing potential bias and that parents would influence the child's responses. Amongst the issues that participants were interested in, which were not addressed in this Delphi, included the views of parents on who should be asked and the adaptation of methods for children in health state valuation.

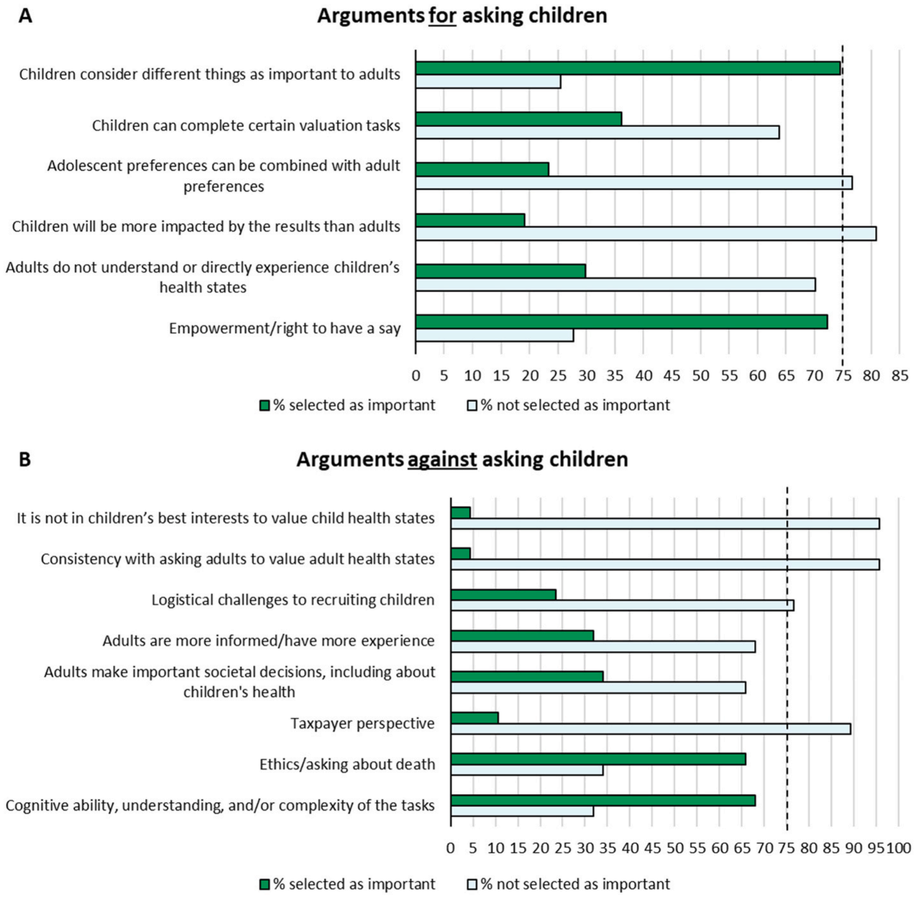

Fig. 3. Percentage of arguments selected as important for asking children to value the health states (A) and against asking children to value the health states (B). Note: Threshold for Delphi consensus set at ≥ 75%. Responses taken from Round 2 of the Delphi survey.

3.2. Who should be imagined?

3.2.1. Round 1

No consensus was observed on who should be imagined (i.e., which perspective) for adults (18+ years). Consensus was achieved for using an 'own' perspective for older adolescents (16–17 years) (77.6%) and younger adolescents (10–15 years) (84.2%) (Fig. 4). Participants found it difficult to provide a specific age that participants should think about if adopting an 'other child' perspective, with the majority refusing to answer or not answering directly. Of the minority that did respond (n = 13–17), the median response was 10 years for all age categories.

Consensus was observed that participants should know they are valuing child health states, regardless of the perspective used (79.3%). No formal consensus was achieved on endorsement **for** taking an 'own' perspective, the most endorsed argument was *it is difficult to imagine for others* (62.3%) and the least endorsed was *veil of ignorance/bias* (27.6%).

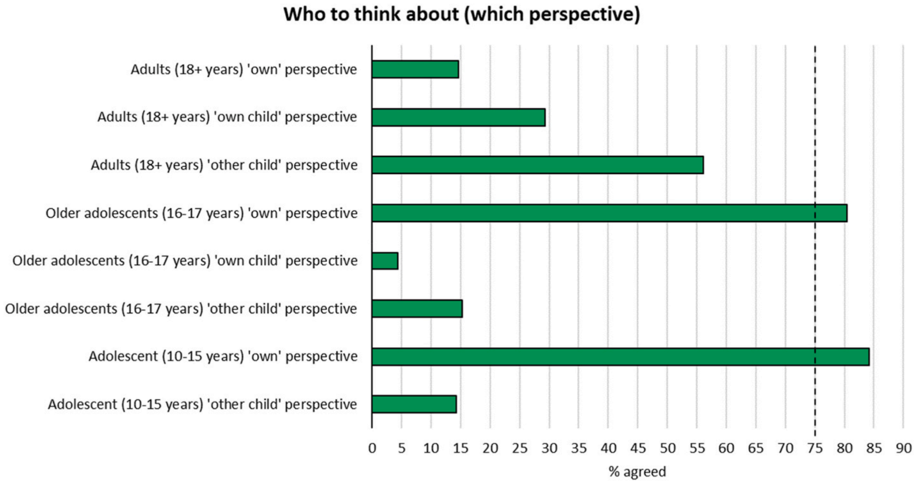

Fig. 4. Percentage agreement for who should be imagined (which perspective).
Note: Threshold for Delphi consensus set at ≥ 75%. Data for adults and older adolescents taken from Round 2 of the Delphi survey, and percentages displayed exclude those who selected the option that this age group 'should not be asked to value child health states'. Data for adolescents (10–15 years) taken from the Round 1 survey.

Regarding arguments **against** taking an ‘own’ perspective, the argument *participants have a right to know it is about children* reached consensus (75.0%), and *consistency with prior research in valuing child health* received the least endorsement (19.3%).

Regarding qualitative findings, mixed views were shared on the use of an ‘own’ vs. ‘other child’ perspective for adults (18+ years) valuing child health, with participants acknowledging problems with both approaches. The vast majority of the fourteen participants who commented for older adolescents (16–17 years) and all ten who commented for younger adolescents (10–15 years) advocated the use of an ‘own’ perspective for these age groups. Qualitative comments on choosing a specific age to imagine in an ‘other child’ perspective were inconsistent, with some participants questioning the basis of picking a particular age, including the current EQ-5D-Y usage of a ‘10-year-old child’.

Qualitative comments on arguments were mixed. Most participants who chose to comment shared critical reflections. For example, participants were critical of the argument *participants have a right to know it is about children* as an argument for the ‘other child’ perspective, as the latter could be achieved with an informed adult ‘own’ perspective. Of the remaining comments, several participants noted that the difference and implications between imaging self as a child versus imagining another child had not been adequately covered. Furthermore, one participant queried whether, as well as knowing the valuation was about child health states, participants should also be informed about how the values could be used (i.e., to inform health resource allocation).

3.2.2. Round 2

Several modifications were made for the Round 2 survey on who to imagine (Supplementary File B). In particular, categorical questions asked about whether adults (18+ years) or older adolescents (16–17 years) should take an ‘own’ perspective; ‘own child’ perspective (i.e., think about themselves as a child); or ‘other child’ perspective when valuing the child health states. An initial error in analysis of Round 1 responses, subsequently corrected, meant that the question on perspective for older adolescents was judged as approaching (rather than reaching) consensus, so this topic was retained in Round 2. Free-

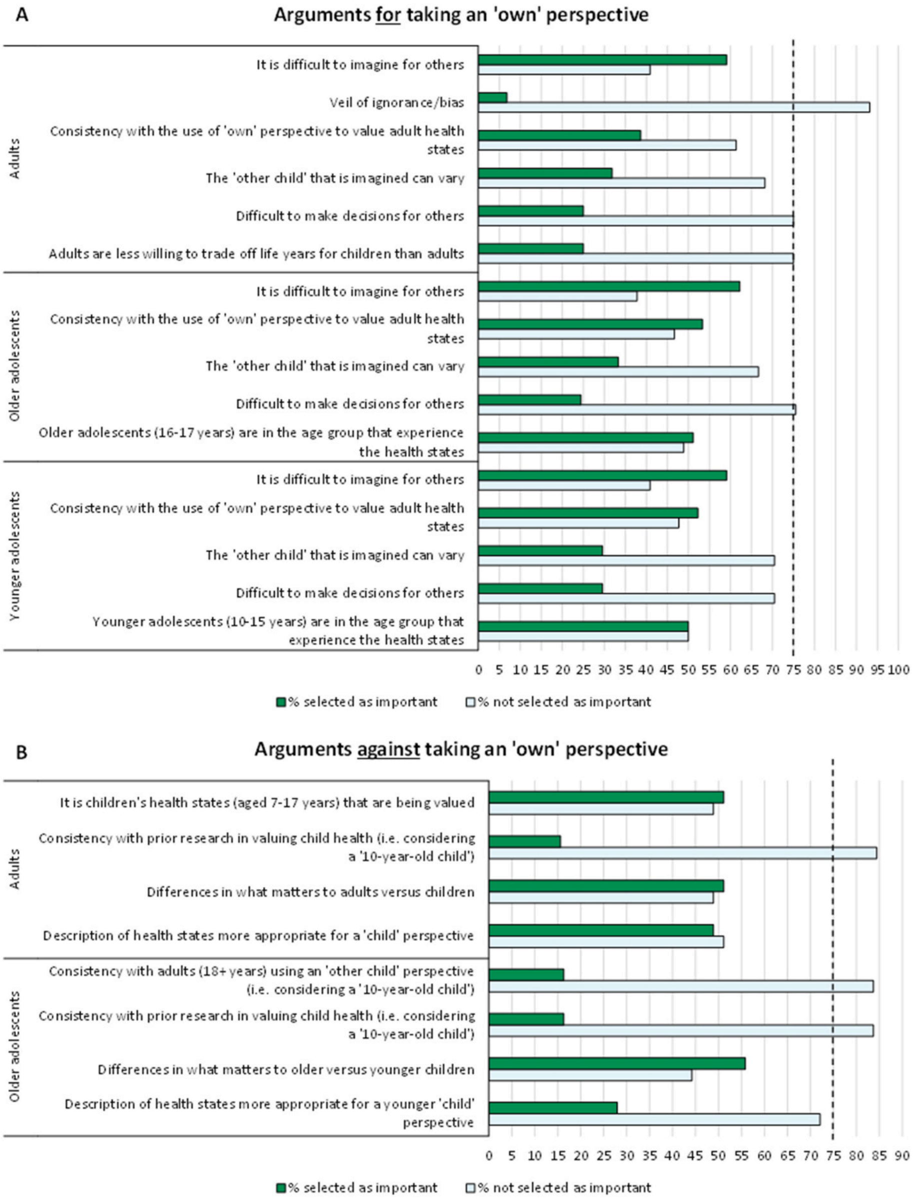

Fig. 5. Percentage of arguments selected as important for taking an ‘own’ perspective (A) and against taking an ‘own’ perspective (B) for different groups valuing the child health states.
Note: Threshold for Delphi consensus set at ≥ 75%. Responses taken from Round 2 of the Delphi survey.

text questions on identifying a specific age child to imagine were deemed too difficult, so instead possible categorical response options were provided (i.e, 10-year-old, 12-year-old, unspecified, or another specified age). Two additional Likert questions were added on: (i) whether the age of the child participants are asked to think about should be based on the age range of children who experience the health state(s); and (ii) whether participants should know that their responses may influence funding decisions in children's healthcare. Finally, arguments were modified in the same way as for who to ask (see above). However, for arguments against an 'own' perspective, where there were four alternatives to choose from, participants were asked to select up to 2 that they thought were most important or persuasive (rather than 3). Questions were asked separately for adults (18+ years) and older adolescents (16–17 years).

For adults (18+ years) valuing the child health states, there was no consensus on exactly who they should imagine (i.e., an 'own' or 'other' child perspective). However, there was a consensus view, amongst those who were not against adults valuing child health states, that they should adopt some kind of 'child' perspective (85.4%). For older adolescents (16–17 years), there was consensus that they should adopt an 'own' perspective when valuing the health states (78.7%) (Fig. 4). As in Round 1, there was no consensus on what age child a participant adopting a 'child' perspective should think about. The idea that participants should be told their responses may indirectly influence funding decisions in children's healthcare approached consensus (72.3%).

As with who to ask, no consensus was observed in the arguments selected as important for who to imagine, but some consensus was seen in those not selected as important (Fig. 5). For example, regarding adult arguments **for** using an 'own' perspective, *veil of ignorance/bias* was not selected as important by 93.2% of participants, and *difficult to make decisions for others* and *adults are less willing to trade off life years for children than adults* was not selected as important by 75% of participants. *Difficult to make decisions for others* was also seen as less important as a reason for older adolescents to take an 'own' perspective (not selected by 75.6%).

For arguments **against** using an 'own' perspective, *consistency with prior research in valuing child health (i.e. considering a '10-year-old child')* was viewed as least important, it was not selected by 84.4% of participants, when considering adults, and 83.7% when considering older adolescents. Similarly, *consistency with adults (18 + years) using an 'other child' perspective (i.e. considering a '10-year-old child')* was not considered an important argument for older adolescents (not selected by 83.7%).

Qualitative comments on the use of an own vs. other perspective in Round 2 were sparse and mixed. Of the participants who commented on specifying a specific age of child to imagine, most identified that the choice was arbitrary and problematic. Some referred to evidence that the age of the child did matter in valuation, while others referred to evidence that said it did not. A few participants commented on the need to adapt the age (range) to fit with the health states being valued. Three participants stated that they chose a 10-year-old (for generic health states) based on the EQ-5D-Y approach. Two participants who commented on the specific age of child to imagine noted a need for consistency between who adults and older adolescents were asked to imagine. Comments on whether people should know their responses could influence resource allocation decisions were mixed. Two participants were in favour of doing so for transparency and ethical reasons, but one of those noted potential bias.

4. Discussion

This Delphi study was designed to establish the degree of consensus on the normative positions of who to ask and who should be imagined when valuing child health in the UK. An international panel was recruited (with UK policymakers) and the response rate was positive and in line with prior norms (Gargon et al., 2019). At the end of two rounds, consensus was observed on some key issues, with some areas approaching consensus, and some without consensus. The survey was ended without a third round as the response rate was dropping and any anticipated further movement in consensus was insufficient to warrant the additional resources required (i.e., a diminishing returns argument (Hasson et al., 2000)). Instead, areas where consensus was not established serve to "provide informative insights and highlight differences in perspectives concerning the topic in question" (Jünger et al., 2017), and represent areas for further development and research.

Regarding who to ask, consensus was observed that both older adolescents and adults (i.e., those ≥16 years) should be included in the sample valuing the child health states. These findings broadly align with the views of a sample of the UK public, who advocated for greater involvement of children of an appropriate age and maturity (Powell et al., 2023) and with emerging qualitative stakeholder research from Canada (Xie et al., 2024). However, this approach differs from the current EQ-5D-Y-3L valuation protocol, which exclusively involves adults (Ramos-Goñi et al., 2020). Further, there are additional methodological considerations when recruiting both adults and older adolescents in the same valuation study and how the findings are produced for policy (e.g., producing separate vs. combined value sets) that did not reach consensus, and thus require further attention (Rowen et al., 2022). Indeed, these findings suggest that stakeholders think that adults and older adolescents should be treated *differently* in the same child health valuation study, with the former taking some form of 'child' perspective and the latter adopting an 'own' perspective. This presents methodological issues for how the responses may or may not be combined and analysed.

A distinction was made throughout the Delphi between valuation methods that involved trading off life years and those that did not (that were anchored to the scale required to generate QALYs by some other method). This distinction is important and valuation tasks that involve having to imagine a reduced lifespan is a common argument given for not involving children in those tasks (Rowen et al., 2020; Ramos-Goñi et al., 2020; Kwon et al., 2022; Kreimeier and Greiner, 2019; Rogers et al., 2021). There was consensus that younger children (i.e., 15-year-olds and above) could be included if the valuation task did not involve trading off life years. Further, participants agreed that children aged ≤10 years old should not be included if the task involved trading off life years. However, in the age ranges between this, further discussion, debate, and research in this area is needed, perhaps involving greater nuance (e.g., type of task and purpose of valuation) and the views of wider stakeholders, such as experts in child development and/or adolescents themselves.

Regarding the arguments underlying the involvement of children and adolescents in health state valuation, *empowerment/right to have a say* was one of the most important (i.e., approaching consensus). This is consistent with the views of members of the UK public (Powell et al., 2023). Of particular interest was that, despite being used to justify asking adults to value child health states (e.g., (Ramos-Goñi et al., 2020)), the *taxpayer perspective* was not selected as an important argument for consideration by the majority of participants. This reinforces the view articulated elsewhere that the taxpayer argument for asking adults to value child health states (when informing health resource allocation) is not a primary consideration (Powell et al., 2023).

Regarding who to imagine (which perspective), there was consensus that adolescents (aged 10–17 years) valuing child health states should take an 'own' perspective, and this is consistent with qualitative findings (Powell et al., 2023; Xie et al., 2024). While consensus emerged that adults should take a 'child' perspective of some form (Powell et al., 2021), there was neither consensus over which form (i.e., 'own child' vs. 'other child' perspective), nor the specific age of child that should be imagined. Reflecting qualitative comments in the survey, researchers have pointed out that the choice (e.g., to choose to imagine a 10-year-old child) is somewhat arbitrary (Lipman et al., 2021b). Mixed evidence is emerging over the extent to which changing the age of the child that participants are asked to imagine impacts valuation and this is an area for further research (Reckers-Droog et al., 2022; Ramos-Goñi et al., 2022).

While some of the arguments for who to imagine were considered consensually less important by participants, *participants have a right to know it is about children* was the only argument to reach consensus as important **against** the use of an 'own' perspective (for adults). However, qualitatively, it was commented that this is possible to achieve while still using an 'own' perspective. Consensus was observed that participants should know it is about children when valuing child health states, and this is consistent with members of the UK public's views that it is ethically prudent and best practice to disclose this information (Powell et al., 2023).

A need for additional research to help inform normative views on who to ask and who to imagine was explicitly noted by several participants. This included further research on the consequences of imagining children of different ages in valuation and taking an 'own child' versus 'other child' perspective. There was interest in understanding the views of parents on who they think should be asked in child valuation (Powell et al., 2021). Further, some participants advocated for research into how valuation methods could be developed or adapted to work with younger children. While responses were mixed, a few participants also saw potential promise in dyadic valuation (i.e., involving children and parents) and this may represent another area for further child valuation research (Powell et al., 2023).

4.1. Strengths and limitations

Strengths of this research include the breadth and size of the sample. There is a relatively small number of people researching and undertaking policy decisions in the field of child health valuation worldwide and so this study represents the views of a significant number of invested stakeholders from 18 countries. The study included wide representation from academics, private consultants, people working in not-for-profit organisations, and government policymaker representatives. Further, this Delphi study is the first of its kind in the field and will serve to further stimulate research and debate to move towards consensus in normative methodological decisions in child health valuation.

Limitations of the current study also need to be disclosed. First, as is standard in a Delphi study, recruitment was purposive and targeted, based on desired expertise (Hasson et al., 2000). While a good response rate was observed, the views reflect a select group of experts and may not generalise to the entire field. Reflecting the field, a critical mass of experts (approximately 42%) had some known affiliation to the EuroQol Group and this may have caused some homogeneity in responding. Further, a minority of participants did not self-report prior experience with child health valuation. However, the majority of these had experience with health valuation per se and were UK policymakers who have a vested interest in policy in this area, and thus were deemed suitable for inclusion. Some attrition was observed between Round 1 and Round 2, but this is consistent with normal amounts for a Delphi study (Gargon et al., 2019).

Second, the work was conducted in a UK HTA context (with UK policymakers), so while the responses are expected to have applicability to other countries where QALYs are used in HTA, they may not replicate exactly in other countries. Further, participants responses may have differed if they were being asked to consider valuation for other uses (i.e., not for informing health resource allocation) or if they were shown different information (i.e., instead of UK public views). The findings are thus a reflection of the questions asked and information provided. Further research addressing additional nuance in the issues of valuing child health could be beneficial.

Third, some isolated critical comments on the Delphi survey included that the survey was long, contained too many questions, and that the questions were difficult to answer. While the study was long, it is not unheard of for Delphi surveys to be of this length, as they tend to be detailed and targeted at experts and offer opportunity for detailed qualitative feedback (Beiderbeck et al., 2021). The findings must be viewed within the context of potential uncertainty around participants' responses. However, participants were able to 'opt-out' of answering questions that they did not feel qualified to answer. Rather than being definitive, this study is meant to represent a current snapshot of consensus and non-consensus in this area, for informing future decision-making and research.

Finally, while up to three rounds were planned, only two Delphi rounds were conducted. This was a decision made by the research team based on the additional resources required to run another round relative to the additional consensus or clarification(s) anticipated. It is possible that consensus (at the level of $\geq$75%) is difficult to achieve on some of the subjective normative issues explored in the Delphi. Further, running additional Delphi rounds when the sample size is dropping is related to a risk of 'false consensus' achieved through sample attrition, rather than changing views (Humphrey-Murto and de Wit, 2019). As the aim of this study was to identify both where consensus does and does not exist, rather than, for example, definitively identifying consensus for the construction of an output (e.g., a reporting checklist), the restriction to two rounds was deemed acceptable.

5. Conclusions

This is the first Delphi study of its kind to attempt to establish degree of expert consensus on the normative issues of who to ask and who to imagine when valuing child health for HTA. The findings have implications both for methodological decisions taken on valuing child health internationally and recommendations made by NICE in the UK context. The research complements ongoing qualitative consultations with the public and other stakeholders for their views on these normative questions and will help inform UK policymakers on the current state-of-play, including where future academic research and/or consensus-building is required.

Key findings from this research suggest that experts think that a combination of older adolescents (16–17 years) and adults (18+ years) should be involved in valuing child health states (aged 7–17 years) and those aged 10 years and under should not, when trading off life years. Experts think that adolescents should take an 'own' perspective and adults should take some form of 'child' perspective. However, there is no consensus about the form that the 'child' perspective for adults should take (e.g., thinking about themselves as a child of a specific age or thinking about another hypothetical child of a specific age) and work is clearly needed to help inform consensus views in this area (including what age child participants should be asked to imagine). Including older adolescents in valuation differs from the currently established EQ-5D-Y-3L valuation protocol (Ramos-Goñi et al., 2020) and methodological choices are evident that require further consideration and research efforts (Rowen et al., 2022). Further research is needed to come to a consensus view on whether younger children should be included in valuation.

Taken together, these results are largely consistent with recent views elicited qualitatively from members of the public and other stakeholders on normative issues in valuing child health (Nazari et al., 2022; Powell et al., 2023; Xie et al., 2024). Efforts should be made to involve older adolescents (16+ years) in child health valuation internationally and specifically in the UK context. Furthermore, research should be conducted to better inform methodological decisions on several outstanding issues, including which form of 'child' perspective adults should take.

Funding

This research is funded by the National Institute for Health and Care Research (NIHR) Policy Research Programme, conducted through the Policy Research Unit in Economic Methods of Evaluation in Health and Social Care Interventions, PR-PRU-1217-20401. The views expressed are those of the author(s) and do not necessarily reflect those of the NHS, the National Institute for Health Research, the National Institute for Health and Care Excellence, the Department of Health and Social Care or its arm's length bodies, or other UK government departments. Any errors are the responsibility of the authors.

Ethics approval

Ethics approval for this study was obtained from the University of Sheffield (reference number: 046269).

CRediT authorship contribution statement

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

Declaration of competing interest

Donna Rowen, Clara Mukuria, and Koonal Shah are members of the EuroQol Group. Koonal Shah is Associate Director at the National Institute for Health and Care Excellence. Philip Powell and Anju Keetharuth have no competing interests to declare.

Data availability

Data will be made available on request.

Acknowledgements

We would like to acknowledge and thank all Delphi participants: Sarah Acaster, Lazaros Andronis, Mimmi Åström, Edward Aveyard, Cate Bailey, Sophie Cooper, Nancy Devlin, Brigitte Essers, Titi Sahidah Fitriana, Wolfgang Greiner, Alison Hayes, Mike Herdman, I-Chan Huang, Paul Jacklin, Michał Jakubczyk, Jennifer Jelsma, Milad Karimi, Simone Kreimeier, Stefan Lipman, Andrew Lloyd, Nan Luo, Louis Matza, David Mott, Christine Mpundu-Kaambwa, Bhash Naidoo, Richard Norman, Marko Ogorevc, Jan Abel Olsen, Tianxin Pan, Becky Pennington, Stavros Petrou, Simon Pickard, Lisa Prosser, Fredrick Dermawan Purba, Benjamin Purchase, Juan M. Ramos-Goñi, Julie Ratcliffe, Vivian Reckers-Droog, Bryce Reeve, Fanni Rencz, Oliver Rivero-Arias, Janet Robertson, Tomos Robinson, Bram Roudijk, Valentina Prevolnik Rupel, Eric Slade, Katie D. Stewart, Elly Stolk, Victoria Thomas, Dominic Thorrington, Aki Tsuchiya, Wendy Ungar, Janine Verstraete, Wang Pei, David Whitehurst, and seven people who wished to remain anonymous. We would also like to thank Allan Wailoo for his advice and input, and Donna Davis for her professional support on this project.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.socscimed.2024.117127.

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