Whose opinion matters when valuing child and adolescent health? Novel insights from surveys of the adult and adolescent public in the UK and Sweden

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Disclaimer

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Abstract

Objective: Normative decisions on who should value child and adolescent health, and who should be imagined when doing so, are important methodological considerations for health technology assessment (HTA) organisations and international research associations such as the EuroQol Group. Consultative evidence from members of the public, whose preferences may be elicited when valuing health, including what adolescents themselves think, can help meaningfully inform such normative decisions. This study aimed to elicit the views of the United Kingdom (UK) and Swedish adult and adolescent public on who they think should be asked to value child and adolescent health.

Methods: An online survey methodology was used. A target sample of 1200 15+ year olds (50/50 split by gender) was recruited in the UK and Sweden. Novel multimedia resources to explain child and adolescent health valuation to the public were developed with expert and public advisory teams, including 15-17 year olds. These resources were embedded into the survey and participants' understanding checked. Survey questions included whether and at what age children should be involved in valuation; whether 15-17 year olds would like to be involved themselves; whether and what prior experience was deemed necessary; and whether trading off life years was acceptable for children, amongst other methodological issues. Data were analysed descriptively, with exploratory comparisons made by country.

Results: The dataset included 1200 UK and 1206 Swedish participants (355 and 363 15-17 year olds, respectively). 59.5% UK and 60.5% Swedish participants thought that both children and adults should value child health. 70.0% UK and 76.5% Swedish participants thought that 16-17 year olds should be asked to value child health, reducing to 18.6% and 20.2%, respectively, for 8-9 year olds. 57.8% UK and 60.9% Swedish participants ($\chi2=14.800(3)$, $p=.002$) expressed that prior experience among adults working with or caring for children was needed to value child health appropriately. 74.9% of 15-17 year olds in the UK and 84.3% in Sweden ($\chi2=10.582(2)$, $p=.005$) wanted to be eligible in principle to value child HRQoL (66.8% and 71.3% when trading off life years). 88.9% UK and 87.2% Swedish participants thought it was acceptable for children and adolescents to trade off life years, though the age at which this was deemed acceptable varied. In both countries, children's

right to have a say was rated as the most important normative argument for who should value child health and the taxpayer argument was rated as the least important.

Conclusion: While some differences were observed in the strength of agreements, modal responses were similar across the UK and Swedish public on most normative issues. Most respondents thought that older children and adolescents should be involved in valuing child and adolescent health and that it would be acceptable for them to trade off life years. The majority of 15-17 year olds wanted to be eligible to value child and adolescent health. In context with other evidence, these findings can help to support normative methodological decisions made when valuing EQ-5D-Y measures and in the EQ-5D-Y-5L valuation protocol.

Introduction

In studies designed to value health states, normative methodological decisions need to be made about the *source* of the values and *perspective* used. Specifically, which group(s) of people are recruited to complete the valuation tasks (e.g., those valuing *described* health states vs. those valuing *experienced* health states), and who they are asked to think about when doing so (e.g., themselves, another person). For adult health states, national recommendations are made by relevant decision-making bodies, such as the National Institute for Health and Care Excellence (NICE) who advocate asking a representative sample of the adult United Kingdom (UK) public to value *described* health states [1], or the Swedish Dental and Pharmaceutical Benefit Agency (TLV) who prefer asking people with experience to value *experienced* health states [2,3]. For child and adolescent health states, however, there are no such recommendations and we are at a time of evolving guidance. Several additional normative and methodological questions are applicable to valuing child and adolescent (versus adult) health, including whether children and adolescents are asked to provide their values; what age ranges and methods are appropriate; and if adults are asked to value the health states, what framing (or 'perspective') should be used.

The first value set developed for the EQ-5D-Y-3L was developed based on a population sample of children and their assessment of their own health state [4], and later the same approach was used by Åström et al. [5]. Recently the focus on developing country-specific value sets for the EQ-5D-Y-3L (e.g., [6-15]) has followed a standardised valuation protocol that has taken an explicit stance on the abovementioned normative issues, stating that the adult general population (18+ years of age) should value health states for a 10-year-old child [16]. This deviates from traditional EQ-5D valuation methodology, by asking adults to value health for someone else (not themselves) and for a child (not an adult). While the 'taxpayer perspective' is cited in this protocol as the justification for using adult members of the public (i.e., they collectively bear the costs of healthcare), no additional rationale is given for the use of a 10-year-old child perspective [17]. Methodological decisions like these can be difficult to make, with arguments existing for and against different approaches, making them tricky to justify without drawing on some external evidence or consultation. Following the logic of the 'taxpayer perspective' (i.e., that the preferences of those who collectively

fund healthcare should be considered), it is argued that a consultation on the public's views about what normative decisions should be made when valuing child health would be beneficial. It is also argued that a consultation with the people whose potential health states the EQ-5D-Y is designed to model (children and adolescents) would also be beneficial.

Historically, health economists have made arguments for and decisions on who should be asked using what perspective when valuing health (see [18]). These decisions have been made with little consultation from those who bear the cost of, and potentially benefit from, health technologies. This somewhat contradicts the sentiment of the 'taxpayer perspective' argument that is used to justify the use of adult general population values and, further, stands in contrast to public and patient involvement and engagement (PPIE) initiatives that prevail in health research and are incorporated as part of health technology assessments (HTA) [1,19]. In the case of children and adolescents, the United Nations Convention on the Rights of the Child stipulates that all children with the capability of forming their own views should have the right to express these views in matters affecting them, with weight given to the age and maturity of the child [20]. NICE's patient and public involvement policy states that children and young people should be involved 'on matters pertaining to NICE's work and that affect children and young people's health and wellbeing' (p. 6, [21]). Accordingly, there is an ethical/moral precedent in consulting not just adults, but, at the very least, older adolescents about the issues of whose opinion matters when valuing health in the context of health resource allocation for children.

While a decision has been made for the EQ-5D-Y-3L valuation protocol to have adults value health states for an imaginary 10-year-old child [16], further empirical and normative work must be conducted to establish the appropriateness and acceptability of this process, to provide feedback to decision-makers, and to inform future approaches. While important prior work to date has provided consistent support for the notion of involving older adolescents as participants when valuing child and adolescent health states [22-24], this has been restricted to relatively small targeted qualitative studies that have not systematically asked larger samples about their views. In summary, large-scale quantitative evidence on who the adult and older adolescent public think should be asked to value child and adolescent health states, and in what form, is missing from the evidence base to help inform

researchers and decision-makers on normative issues in valuing health for children and adolescents.

In obtaining such evidence, a further question of interest arises in exploring adult and older adolescent public responses from European countries with different decision-making contexts. Specifically, comparing responses in the UK (where *described* health states are preferred) and Sweden (where *experienced* health states are preferred). Between-country comparisons can help to ascertain whether public views are similar between the two countries or differ, for example, in alignment with the approaches taken by their respective HTA bodies. This approach, with advisory input from and direct feedback to decision-maker representatives from NICE and TLV, is likely to be informative for stakeholders in the HTA process, and may help incrementally contribute to understanding areas of agreement or discrepancies in public perspectives across the two countries.

Aim

The aim of this research project was to elicit the views of the adult (18+ years old) and older adolescent (15-17 years old) public (in the UK and Sweden) on who they think should be asked to value child and adolescent health states, and in what form.

Methods

Development of explanatory resources

Prior to survey design, the first stage of this project focused on the development, piloting and validation (in English and Swedish) of a novel set of multimedia resources designed to explain in lay terms: health state valuation and its implications; the normative issues of child and adolescent health state valuation; the current (EQ-5D-Y-3L) approach; and arguments given for different approaches on who to ask and who to think about when valuing child and adolescent health. Two videos were produced (one on conceptual issues and one on arguments for different positions) and piloted in English through focus groups with EuroQol representatives (n=5), independent experts (n=4), and with Patient and Public Involvement representatives, including 15-17 year old adolescents (n=14). Following refinement, the video resources were translated into Swedish and piloted with adult and adolescent

members of the public (n=9) to ensure understanding. The lay-friendly explanatory videos were embedded in the survey to aid understanding.

Sample and recruitment

A target sample of 1200 participants in each country was established, stratified by age (older adolescents, 15-17 year olds; emerging adults, 18-20 year olds; and established adults, 21+ years old) and gender (50/50 split), with an expected cell size of n=200 in each age/gender group. Participants were recruited through a market research agency panel. While we acknowledge that selection biases exist when recruiting via market research panels, it is an efficient and relatively cost-effective way to get large sample data from the general public and to provide initial insights into the questions posed in this study.

Survey and procedure

Figure 1 shows the progression of the study. A UK (English) version of the survey was drafted by the research team and feedback on the design was elicited from three collaborating groups, including EuroQol representatives; experts in the field independent of the EuroQol Group; and members of the public (including 15-17 year old adolescents). Collaborators were invited to view (and complete) a draft version of the survey and separate focus groups were held to elicit their feedback. The survey was refined based on their feedback, with particular attention given to ensure that the survey was understood and that none of the questions or embedded resources were 'leading'. The survey was translated into Swedish by the research team and three focus groups were held with adult and adolescent members of the public to ensure understanding. Prior to the main survey launch, the UK survey was piloted online using an online panel Prolific (https://www.prolific.com) with 100 adults (18+ years old) and the data checked.

The surveys were hosted on Qualtrics. An English copy is available at: https://shef.qualtrics.com/jfe/form/SV_8HyN4RycDQe0pwi. The context of the survey was defined as valuing changes in children and adolescents' quality of life (aged 8-17 years). "Quality of life" was chosen as it matched the term used in the explanatory resources produced earlier in the project and was deemed easier to understand for members of the public than "health" (which usually has a more restrictive definition).

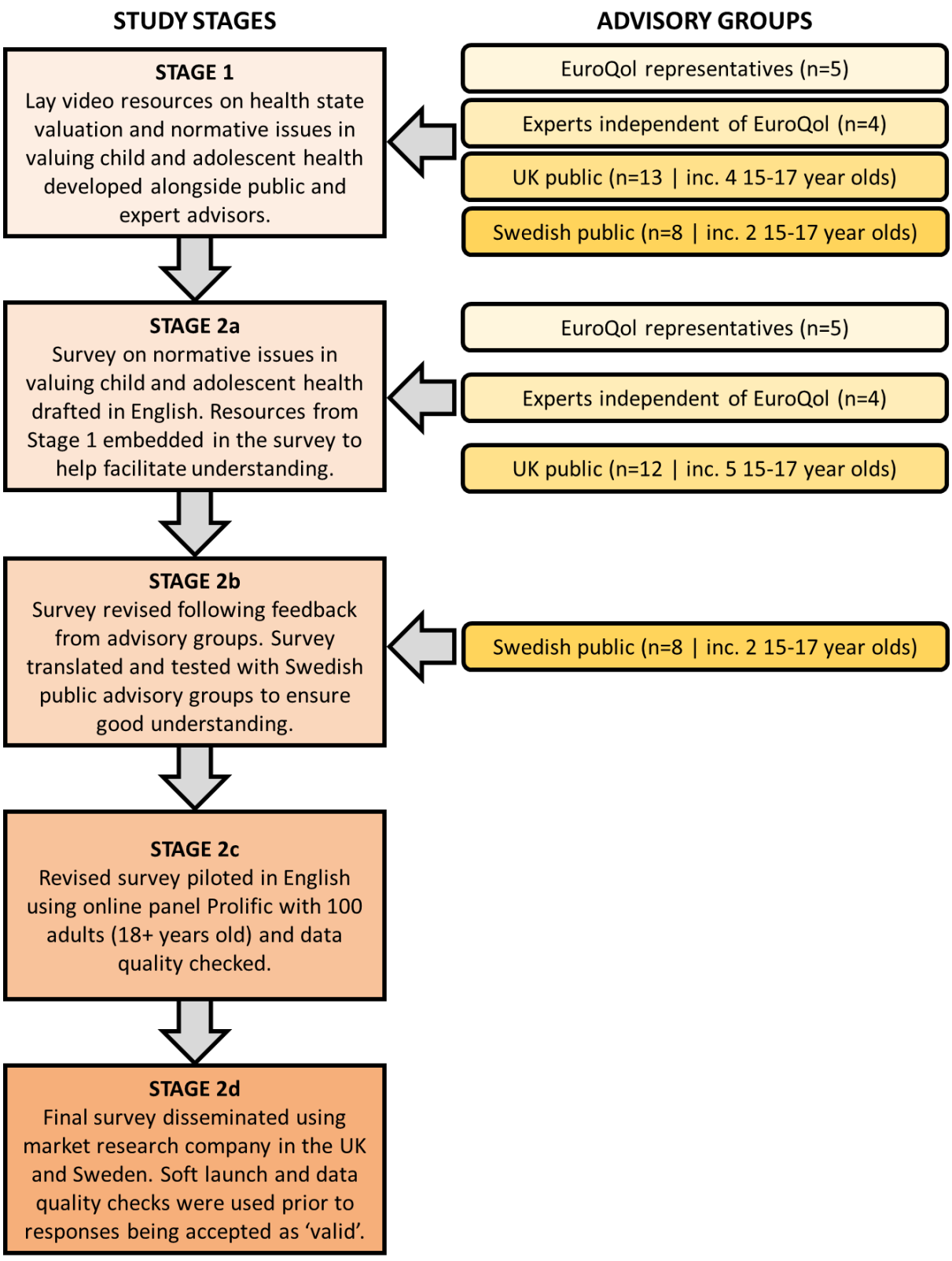

Figure 1. Flowchart of survey study.

Informed consent was taken from the participant (16+ years) or their parents (15 years old). Participants then viewed the first video (on conceptual issues in valuing child and adolescent

health) in full and could not continue with the survey questions until they had finished watching the video. A previous step ensured their audio was working (they were asked to type out what was said in an audio clip to proceed). Three multiple-choice 'video check' questions were included after the first video that assessed participants' understanding of the topic and issues under investigation. As a data quality check, participants who got all three questions incorrect were screened out of the survey. Those who got one or two wrong received feedback on why the answer(s) was wrong and received further written clarification on the key issues that the video and survey were addressing.

After the video, and following initial questions on sociodemographics, questions included whether and at what age children should be involved in valuation; whether 15-17 year olds would like to be involved themselves; whether and what prior experience was deemed necessary; and whether trading off life years was acceptable for children, amongst other methodological issues. Participants were also asked to judge the perceived importance of a range of arguments that have been provided in the literature for and against different perspectives and source of values (see Helgesson et al. for a review [18]) on a 4-point Likert scale (1 = 'Not at all important'; 4 = 'Extremely important'). The second video explaining arguments for and against different positions was embedded before the questions asked on the perceived importance of different arguments. At the end of the survey participants completed the EQ-5D-5L for themselves. Presentation of the arguments for and against different positions was counterbalanced, so that 50% of participants were provided with the questions on arguments before the main questions on who to ask/who to think about and 50% afterwards. Initial analyses suggest presentation order of the arguments had no systematic influence on response patterns.

Data received from the market research panel were screened for data quality before being accepted. Data were screened for duplicate responses (same internet protocol (IP) address, gender, and age), speeding (<50% of the median; [25,26]), unusual response patterns (e.g., obvious straight-lining of responses throughout the survey), and duplicate or suspicious free text responses, and any responses failing data quality checks were rejected.

Ethics approval was granted from the University of Sheffield (reference: 056699).

Data analysis

Quantitative response data were analysed descriptively and reported as frequency (%) or measures of central tendency (i.e., means and standard deviations). Exploratory comparisons by country (i.e., chi-square tests or t-tests) were conducted on R v4.2.2. All comparisons were exploratory in nature and not intended to confirm differences in preferences between countries. No adjustment for multiple testing was made. Due to the volume of free text (i.e., qualitative) responses, they are not analysed or presented in this discussion paper, but will be explored in future work.

Results

Participants

A total of 1200 participants from the UK and 1206 from Sweden completed the survey. Sociodemographic background characteristics are outlined in Table 1. The UK and Swedish samples were broadly comparable on many criteria, including gender and age, presence of a degree, and parental status. However, the samples differed in some ways: including employment status (i.e., more unemployed in the UK sample vs. more participants who were students/in training in the Swedish sample), health (i.e., the UK sample self-reported better health than the Swedish sample), and experience working with children (i.e., more participants in the Swedish data reported having worked with children in a professional or voluntary capacity). A total of 718 15-17 year olds were recruited (355 in the UK and 363 in Sweden). Their details are summarised separately in Table 1.

Table 1. Participants' sociodemographics

	Full sample (2406)		**χ^2 or t**	**15-17 year olds (718)**		**χ^2 or t**
	N (%) or M (SD)			**N (%) or M (SD)**		
	UK	**Sweden**		**UK**	**Sweden**	
Gender			1.075(3), p = 0.783			0.040(3), p = 0.998
Woman	603 (50.3)	601 (49.8)		182 (51.3)	183 (50.4)	
Man	573 (47.8)	583 (48.3)		168 (47.3)	174 (47.9)	
Non-binary	15 (1.3)	15 (1.2)		4 (1.1)	4 (1.1)	

Other	3 (0.3)	1 (0.1)		1 (0.3)	1 (0.3)	
Age (years)	26.94 (15.57)	26.79 (15.14)	−0.235 (2305.5), p = 0.815	15.97 (0.82)	15.99 (0.85)	0.455(715.73), p = 0.649
Ethnicity[a]			N/A			N/A
Asian or Asian British	138 (11.5)	/		39 (11.0)	/	
Black, Black British, Caribbean or African	157 (13.1)	/		47 (13.2)	/	
Mixed or multiple ethnic groups	56 (4.7)	/		26 (7.3)	/	
White	817 (68.1)	/		233 (65.6)	/	
Other ethnic group	16 (1.3)	/		8 (2.3)	/	
English/Swedish as first language			0.466(1), p = 0.495			0.040(1), p = 0.842
Yes	1093 (91.1)	1088 (90.2)		328 (92.4)	332 (91.5)	
No	100 (8.3)	111 (9.2)		27 (7.6)	30 (8.3)	
Bachelors or higher degree qualification[b]			1.042(1), p = 0.307			N/A
Yes	246 (31.0)	227 (28.4)		/	/	
No	546 (68.8)	567 (71.0)		/	/	
Primary employment status[c]			25.218(4), p < 0.001			42.026(3), p < 0.001
Student/in training	385 (37.6)	493 (47.9)		178 (77.1)	223 (96.5)	
Employed	438 (42.7)	393 (38.2)		22 (9.5)	4 (1.7)	
Unemployed[d]	142 (13.9)	97 (9.4)		29 (12.6)	2 (0.9)	
Retired	47 (4.6)	40 (3.9)		0 (0.0)	0 (0.0)	
Other[e]	5 (0.5)	3 (0.3)		0 (0.0)	1 (0.4)	
Overall health			18.743(4), p < 0.001			8.189(4), p = 0.085
Very good	262 (21.8)	235 (19.5)		98 (27.6)	77 (21.2)	
Good	635 (52.9)	585 (48.5)		187 (52.7)	215 (59.2)	
Neither good nor bad	212 (17.7)	247 (20.5)		55 (15.5)	48 (13.2)	
Bad	68 (5.7)	109 (9.0)		12 (3.4)	19 (5.2)	
Very bad	14 (1.2)	25 (2.1)		1 (0.3)	4 (1.1)	

Day-to-day activities limited by health			7.819(2), p = 0.020			0.966(2), p = 0.617
Yes, limited a lot	110 (9.2)	108 (9.0)		18 (5.1)	24 (6.6)	
Yes, limited a little	257 (21.4)	319 (26.5)		62 (17.5)	68 (18.9)	
No	806 (67.2)	762 (63.2)		269 (75.8)	268 (73.8)	
EQ-5D-5L utility[f,g]	0.891 (0.192)	0.883 (0.187)	−1.066 (2369.7), p = 0.287	0.921 (0.145)	0.918 (0.156)	−0.228 (705.67), p = 0.820
EQ-5D VAS[h]	76.29 (20.37)	74.56 (21.06)	−2.028 (2367.4), p = 0.043	80.88 (17.00)	80.64 (17.49)	−0.180 (703.92), p = 0.857
Parent or guardian[c]			0.986(1), p = 0.321			1.145(1), p = 0.285
Yes	255 (24.9)	236 (22.9)		6 (2.6)	2 (0.9)	
No	770 (75.1)	794 (77.1)		225 (97.4)	229 (99.1)	
Child under 18 years old[i]			0.066(1), p = 0.797			N/A
Yes	156 (61.2)	148 (62.7)		6 (100.0)	2 (100.0)	
No	99 (38.8)	88 (37.3)		0 (0.0)	0 (0.0)	
Child day-to-day activities limited by health[i]			2.410(2), p = 0.300			2.667(2), p = 0.264
Yes, limited a lot	19 (7.45)	24 (10.2)		1 (16.7)	1 (50.0)	
Yes, limited a little	43 (16.9)	48 (20.3)		4 (66.7)	0 (0.0)	
No	191 (74.9)	163 (69.1)		1 (16.7)	1 (50.0)	
Ever worked with children[b]			4.344(1), p = 0.037			NA
Yes	306 (38.5)	350 (43.8)		/	/	
No	488 (61.5)	449 (56.2)		/	/	

Note. Sample may not add up to 100% because of 'prefer not to say' responses. [a]Ethnicity question was not asked in Sweden due to cultural conventions. [b]Sample restricted to 18+ year olds (n=1593). [c]Sample restricted to 16+ year olds (n=2055 (462 16-17 year olds)). [d]Includes job seekers, people unemployed due to disability or sickness, and people not seeking work. [e]'Other' responses included full-time carers and/or people who self-defined as home-makers. [f]Calculated using the Swedish value set by Sun et al. (2022) [27]. [g]Missing data on the EQ-5D-5L (n=20 missing in UK and n=11 in Sweden). [h]Missing data on the EQ-5D VAS (n=25 missing in UK and n=11 in Sweden). [i]Sample restricted to parents or guardians (n=491 (8 16-17 year olds)).

Data overview

The median time to complete the survey was 19.4 minutes for the UK version and 23.0 minutes for the Sweden version. 77.7% in the UK sample and 83.0% in the Sweden sample answered at least two of the three 'video check' questions correctly (15.6% likely to happen by chance). 83.3% of UK participants and 71.6% of Swedish participants affirmed that when they completed the survey they were "aware that information on the value of changes in children and adolescents' quality of life is used to inform public healthcare funding decisions." 79.8% of participants in the UK and 77.7% in Sweden reported being "quite" or "very" certain about their responses when answering the survey. 38.5% of UK responses and 39.6% of Sweden responses featured at least one "I don't know" response (with 18.3% and 19.2% featuring only a single "I don't know" response, respectively).

Regarding questions on understanding, 61.8% of the UK sample and 59.5% of the Sweden sample reported that the topic and questions on who to ask were "quite" or "very" easy to understand (a further 21.4% and 24.3% reported that they were "neither difficult nor easy" to understand, respectively). The respective percentages for ease of understanding the topic and questions on who to think about (which perspective) were 55.3% and 51.8% (with 25.3% and 27.9% reporting that it was "neither difficult nor easy"). 17.2% in the UK and 18.0% in Sweden reported finding the questions on who to ask difficult to answer, with 21.5% and 23.0%, from each respective country, reporting finding the questions on who to think about difficult to answer.

The results below are reported for the entire sample (n=2406) and for the subsample of adolescents aged 15-17 years (n=718), who represent a unique group of interest. Further sensitivity analyses with participants removed who failed aspects of the quality control criteria above and/or had inconsistencies in their responding will be possible in a future version of this paper.

Who should be asked to value child and adolescent health?

Table 2 shows the results on opinions on who to ask to value child and adolescent health. The majority of participants in both the UK and Sweden (59.5% and 60.5%, respectively) supported asking both children and adolescents to value child and adolescent health. This

was also true of the subsample of older adolescents themselves. Where children and adolescents were to be involved in valuation, a majority of participants supported asking 14-15 year olds (58.4% and 66.7%) and 16-17 year olds (70.0% and 76.5%), with a pattern of a greater level of support for inclusion of these age groups in Sweden across the full sample.

A majority of participants in both countries endorsed the idea that people valuing child and adolescent health should have experience of ill health (50.1% and 52.2% in the UK and Sweden, respectively) and with children (57.8% and 60.9%, respectively), with significantly greater support for the latter in the Swedish sample. A similar pattern was observed in 15-17 year olds, when considered separately. What participants thought of as potentially 'relevant experience' of ill health and with children varied (see Table 2).

Almost all participants thought that children and adolescents could take part in tasks that involved thinking their age of death and shortening their years of life, but responses were split as to whether this would be appropriate for all children and adolescents (45.7% and 43.3% for the UK and Sweden, respectively), or only those over a certain age (43.2% and 43.9%, respectively). The 15-17 year old subsample were more likely to say that this was appropriate for all 8-17 year olds (50.1% and 52.6% in the UK and Sweden, respectively). For those who said that this was only appropriate for older adolescents, the median minimum age for taking part in such tasks in principle was given as 14 and 15 years, by UK and Swedish participants, respectively. Of 15-17 year olds included in the survey, a majority in both countries wanted to be eligible in principle to value child and adolescent health (74.9% and 84.3% in the UK and Sweden, respectively), with a greater desire for participation in the Swedish sample. Of these, 89.1% and 84.6%, in the UK and Sweden, still wanted to be eligible when informed that the tasks involved imagining their age of death (66.8% and 71.3% of the total 15-17 year old sample in each country, respectively).

The argument rated as most important for informing who should be asked to judge the value of changes in children and adolescents' quality of life, in both countries, was 'Children and adolescents have a right to have a say in things that affect them' (combined M = 3.45, SD = 0.74). The argument rated as least important, in both countries, was 'Adults pay taxes and taxes fund the healthcare system' (combined M = 2.60, SD = 0.98; $t(2405) = 34.531$, $p < .001$). This pattern of responses was the same in the 15-17 year old subsample.

Table 2. Participant responses to who should be asked to value child and adolescent health

	Full sample (2406)		χ^2 or t	15-17 year olds (718)		χ^2 or t
	N (%) or M (SD)			N (%) or M (SD)		
	UK	Sweden		UK	Sweden	
Who should value child and adolescent quality of life?						
Adults (aged 18+ years)	223 (18.6)	179 (14.8)	13.050(3), p = 0.005	31 (8.7)	36 (9.9)	4.269(3), p = 0.234
Children and adolescents (aged 8-17 years)	228 (19.0)	276 (22.9)		98 (27.6)	104 (28.7)	
Both adults and children and adolescents	714 (59.5)	730 (60.5)		216 (60.8)	220 (60.6)	
I don't know	35 (2.9)	21 (1.7)		10 (2.8)	3 (0.8)	
If children and adolescents are asked to value child and adolescents' quality of life, which age groups should be asked?[a]						
16-17 year olds	840 (70.0)	922 (76.5)	12.443(1), p < 0.001	273 (76.9)	260 (71.6)	2.344(1), p = 0.126
14-15 year olds	701 (58.4)	804 (66.7)	17.127(1), p < 0.001	242 (68.2)	221 (60.9)	3.850(1), p = 0.050
12-13 year olds	501 (41.8)	577 (47.8)	8.788(1), p = 0.003	167 (47.0)	156 (43.0)	1.041(1), p = 0.308
10-11 year olds	302 (25.2)	349 (28.9)	4.147(1), p = 0.042	99 (27.9)	78 (21.5)	3.620(1), p = 0.057
8-9 year olds	223 (18.6)	244 (20.2)	0.943(1), p = 0.332	62 (17.5)	54 (14.9)	0.707(1), p = 0.400
Do not ask children of any age	145 (12.1)	158 (13.1)	0.477(1), p = 0.490	26 (7.3)	73 (20.1)	23.620(1), p < 0.001
I don't know	73 (6.1)	30 (2.5)	18.112(1), p < 0.001	16 (4.5)	5 (1.4)	5.138(1), p = 0.023
Should people who are asked to value child and adolescents' quality of life have experience of ill health themselves?						
Yes	601 (50.1)	630 (52.2)	1.179(2), p = 0.555	183 (51.5)	192 (52.9)	4.776(2), p = 0.0918
No	392 (32.7)	373 (30.9)		107 (30.1)	125 (34.4)	
I don't know	207 (17.3)	203 (16.8)		65 (18.3)	46 (12.7)	

What counts as relevant experience of ill health?[a,b]						
Own experience of relevant health problems	493 (82.0)	512 (81.3)	0.073(1), p = 0.787	148 (80.9)	160 (83.3)	0.237(1), p = 0.627
Working with/caring for people with relevant health problems	398 (66.2)	444 (70.5)	2.381(1), p = 0.123	124 (67.8)	144 (75.0)	2.067(1), p = 0.151
Having a friend or family member with relevant health problems	316 (52.6)	323 (51.3)	0.162(1), p = 0.687	105 (57.4)	94 (49.0)	2.339(1), p = 0.126
Other	4 (0.7)	8 (1.3)	0.622(1), p = 0.430	2 (1.1)	2 (1.0)	0.000(1), p = 1.000
None of the above	5 (0.8)	4 (0.6)	0.005(1), p = 0.943	2 (1.1)	0 (0.0)	0.552(1), p = 0.457
I don't know	9 (1.5)	11 (1.7)	0.014(1), p = 0.905	3 (1.6)	2 (1.0)	0.003(1), p = 0.957
Should people who are asked to value child and adolescents quality of life have experience caring for or working with children or adolescents?						
Yes	693 (57.8)	735 (60.9)	14.800(3), p = 0.002	203 (57.2)	238 (65.6)	6.53(3), p = 0.088
No	280 (23.3)	257 (21.3)		81 (22.8)	68 (18.7)	
Adults should not value child and adolescent quality of life	100 (8.3)	60 (5.0)		33 (9.3)	21 (5.8)	
I don't know	127 (10.6)	154 (12.8)		38 (10.7)	36 (9.9)	
What counts as relevant experience caring for/working with children or adolescents?[a,c]						
Experience as a parent or guardian	534 (77.1)	602 (81.9)	4.861(1), p = 0.027	160 (78.8)	210 (88.2)	6.513(1), p = 0.011
Working with or looking after children or adolescents (not as a parent or guardian)	572 (82.5)	623 (84.8)	1.132(1), p = 0.287	166 (81.8)	200 (84.0)	0.253(1), p = 0.615
Having relatives that are children or adolescents	316 (45.6)	233 (31.7)	28.530(1), p < 0.001	99 (48.8)	77 (32.4)	11.635(1), p < 0.001
Other	8 (1.2)	11 (1.5)	0.111(1), p = 0.739	2 (1.0)	2 (0.8)	0.000(1), p = 1.000
None of the above	3 (0.4)	1 (0.1)	0.313(1), p	0 (0.0)	0 (0.0)	NA

			= 0.576			
I don't know	9 (1.3)	5 (0.7)	0.840(1), p = 0.359	2 (1.0)	0 (0.0)	0.679(1), p = 0.410
Do you think it is acceptable for children or adolescents to take part in research tasks that involve thinking about their age of death and shortening their years of life?						
Yes, all 8-17 year olds could take part in these tasks	548 (45.7)	522 (43.3)	6.366(3), p = 0.095	178 (50.1)	191 (52.6)	1.617(3), p = 0.656
Yes, for older children and adolescents but not for younger children	518 (43.2)	529 (43.9)		149 (42.0)	149 (41.0)	
No	54 (4.5)	81 (6.7)		10 (2.8)	11 (3.0)	
I don't know	80 (6.7)	74 (6.1)		18 (5.1)	12 (3.3)	
If children and adolescents take part in hypothetical research tasks that involve thinking about their age of death and shortening their years of life, the minimum age for participation (in years) should be:[d,e]						
Mean (SD)	13.79 (2.29)	14.49 (2.14)	5.064 (1036.4), p < 0.001	13.39 (2.27)	14.15 (2.05)	3.025 (292.81), p = 0.003
As a 15-17 year old, would you like to be eligible in principle to value child and adolescent quality of life?[f]						
Yes	/	/	NA	266 (74.9)	306 (84.3)	10.582(2), p = 0.005
No	/	/		35 (9.9)	18 (5.0)	
I don't know	/	/		54 (15.2)	39 (10.7)	
As a 15-17 year old, would you still like to be eligible if the research involved imagining your age of death?[f,g]						
Yes	/	/	NA	237 (89.1)	259 (84.6)	2.754(2), p = 0.252
No	/	/		13 (4.9)	18 (5.9)	
I don't know	/	/		16 (6.0)	29 (9.5)	

Question text paraphrased from the online survey. [a]Answers are not mutually exclusive. [b]Only asked of people that said 'Yes' to 'Should people have experience of ill health themselves?' [c]Only asked of people that said 'Yes' to 'Should people have experience caring for or working with children or adolescents?' [d]Only asked of people that said 'Yes, for older children and adolescents but not for younger children' to 'Do you think it is acceptable for children or adolescents to take part in research tasks that involve thinking about their age of death and shortening their years of life?' [e]Variable censored at minimum 8 years old. [f]Only asked of people who reported their age as between 15 to 17 years. [g]Only asked of people that said 'Yes' to 'As a 15-17 year old, would you like to be eligible in principle to value changes in children and adolescents' quality of life?'

Which perspective should be used?

Table 3 shows the survey results on opinions on who should be thought about (which perspective) when asking people to value child and adolescent health. A majority of participants in the UK and Sweden endorsed the idea of adults (aged 18+ years) imagining a child or adolescent when valuing their health (51.3% and 53.4%, respectively). When adolescents (aged 15-17 years) were valuing health, the majority of participants supported them thinking about themselves, with stronger support for this in Sweden than the UK (52.3% and 44.7%, respectively). This pattern was broadly mirrored for the 15-17 year old subsample. However, Swedish 15-17 year olds were split on the idea of adults imagining themselves vs. imagining a child when valuing child health (44.1% each).

Participants' views on how difficult it would be for them to imagine the importance of changes in quality of life for a 10-year old child were varied and reasonably balanced (see Table 3). A majority of participants in the UK and Sweden thought that they would give different answers for a 15-year-old (63.4% and 75.1%, respectively) and 4-year-old (67.8% and 79.9%, respectively) versus a 10-year-old, with stronger agreement in the Swedish sample. The same pattern of results was reflected in the 15-17 year old subsample.

A majority of participants in both countries agreed that participants in valuation tasks should know that the task is about children and adolescents (82.3% and 88.3% in the UK and Sweden, respectively) and a slightly smaller majority agreed they should know that their responses may influence healthcare funding decisions in children and adolescents (73.0% and 74.0%, respectively). The same pattern of results was reflected in the 15-17 year old subsample.

The argument rated as most important for informing who people should think about when judging the value of changes in children and adolescents' quality of life, in both countries, was 'There are differences in what matters to adults, older adolescents, and younger children' (combined $M = 3.23$, $SD = 0.80$). The argument rated as least important in the UK was 'Difficult to imagine the importance of aspects of quality of life for someone else' ($M = 2.92$, $SD = 0.80$; $t(1199) = 10.564$, $p < .001$). In Sweden, the least important was 'Who people imagine can vary, therefore it is more consistent to ask people to think about themselves'

(M = 2.86, SD = 0.76; t(1205) = 12.580, p < .001). This pattern of responses was the same in the 15-17 year old subsample.

Table 3. Participant responses on perspective when valuing child and adolescent health

	Full sample (2406)		χ^2	15-17 year olds (718)		χ^2
	N (%)			N (%)		
	UK	Sweden		UK	Sweden	
Which perspective should adults (aged 18+ years) use?						
Imagine themselves experiencing that change in quality of life	406 (33.8)	392 (32.5)	1.268(3), p = 0.737	136 (38.3)	160 (44.1)	3.028(3), p = 0.387
Imagine a child or adolescent experiencing that change in quality of life	615 (51.3)	644 (53.4)		171 (48.2)	160 (44.1)	
Adults should not be asked to value children and adolescents' quality of life	92 (7.7)	84 (7.0)		22 (6.2)	23 (6.3)	
I don't know	87 (7.3)	86 (7.1)		26 (7.3)	20 (5.5)	
Which perspective should older adolescents (15-17 years) use?						
Imagine themselves experiencing that change in quality of life	536 (44.7)	631 (52.3)	20.791(3), p < 0.001	181 (51.0)	225 (62.0)	13.485(3), p = 0.004
Imagine a younger child or adolescent experiencing that change in quality of life	463 (38.6)	407 (33.7)		122 (34.4)	111 (30.6)	
Older adolescents should not be asked to value children and adolescents' quality of life	119 (9.9)	77 (6.4)		29 (8.2)	13 (3.6)	
I don't know	82 (6.8)	91 (7.5)		23 (6.5)	14 (3.9)	
How easy or difficult would it be for you to imagine the importance of changes in quality of life for a 10-year-old child?						

Very difficult	65 (5.4)	63 (5.2)	8.975(5), p = 0.110	10 (2.8)	15 (4.1)	1.480(5), p = 0.915
Quite difficult	349 (29.1)	408 (33.8)		94 (26.5)	102 (28.1)	
Neither difficult nor easy	324 (27.0)	318 (26.4)		100 (28.2)	96 (26.4)	
Quite easy	333 (27.8)	316 (26.2)		118 (33.2)	120 (33.1)	
Very easy	86 (7.2)	63 (5.2)		23 (6.5)	21 (5.8)	
I don't know	43 (3.6)	38 (3.2)		10 (2.8)	9 (2.5)	
Would your answers be different for a 15-year-old instead of a 10-year-old?						
Yes	761 (63.4)	906 (75.1)	44.471(3), p < 0.001	232 (65.4)	291 (80.2)	25.977(3), p < 0.001
No	270 (22.5)	160 (13.3)		82 (23.1)	35 (9.6)	
It depends[a]	33 (2.8)	34 (2.8)		13 (3.7)	14 (3.9)	
I don't know	136 (11.3)	106 (8.8)		28 (7.9)	23 (6.3)	
Would your answers be different for a 4-year-old instead of a 10-year-old?						
Yes	813 (67.8)	964 (79.9)	49.736(3), p < 0.001	242 (68.2)	305 (84.0)	27.241(3), p < 0.001
No	252 (21.0)	143 (11.9)		82 (23.1)	35 (9.6)	
It depends[a]	16 (1.3)	17 (1.4)		7 (2.0)	5 (1.4)	
I don't know	119 (9.9)	82 (6.8)		24 (6.8)	18 (5.0)	
Should participants in valuation tasks know that the task is about children and adolescents?						
Yes	987 (82.3)	1065 (88.3)	18.356(2), p < 0.001	298 (83.9)	331 (91.2)	9.266(2), p = 0.010
No	128 (10.7)	78 (6.5)		37 (10.4)	18 (5.0)	
I don't know	85 (7.1)	63 (5.2)		20 (5.6)	14 (3.9)	
Should participants in valuation tasks know that their responses may influence healthcare funding decisions in children and adolescents?						
Yes	876 (73.0)	892 (74.0)	0.506(2), p = 0.776	253 (71.3)	293 (80.7)	8.965(2), p = 0.011
No	233 (19.4)	231 (19.2)		80 (22.5)	53 (14.6)	
I don't know	91 (7.6)	83 (6.9)		22 (6.2)	17 (4.7)	

Question text paraphrased from the online survey. [a]Participants were asked to clarify in a subsequent free-text response (not reported here).

Discussion

To the best of our knowledge, this is the first quantitative survey study exploring members of the public's opinion on who should be asked to value child and adolescent health in the UK and Sweden. Moreover, it is the first study that has elicited the views of older adolescents (15-17 years) directly on this issue. The findings show that, while there were some slight differences in the patterns of the two sample's responses, majority viewpoints in the UK and Sweden were broadly aligned. In particular, there was modal support for involving both children and adolescents in the valuation of child and adolescents' quality of life, with more support for including older rather than younger adolescents (i.e., from 14 years and over). For who to think about when completing tasks valuing child and adolescent health, modal support was for adults (aged 18+ years) thinking about a child or adolescent and for older adolescents (aged 15-17 years) thinking about themselves. The findings on involving adolescents in valuation echo and reinforce those of previous, smaller, qualitative studies with members of the UK public [22,23] and are consistent with focus group studies of expert stakeholders in the US and Canada [28,29], where the involvement of young people as participants in valuation studies was supported. Further, the findings are broadly consistent with an emerging Delphi study of experts in valuing child and adolescent health, including involving older adolescents as participants and the perspective to be adopted [30].

One argument that is typically levied against the involvement of adolescents as participants in valuation studies is the "possible ethical issues associated with the consideration of dead by a sample of children" [16]. However, in the current study almost all participants thought that children and adolescents (of a certain age) could value health, even if the task involved thinking about their age of death and shortening their years of life. Further, a majority of the 15-17 year olds involved in this study wanted in principle to be eligible to be involved, even when fully informed that the task involved imagining their age of death. While ethical considerations do need to be taken into account [31], these findings suggest that the issue of older adolescents imagining a hypothetical age of death may have been overstated by researchers who have not previously consulted with members of the public, including older adolescents, who are the target population for adolescent health state valuation tasks.

A key difference between HTA processes in the UK and Sweden is the use of hypothetical versus experienced values [18]. One may thus expect differences between samples from the

two countries in the prevailing views about whether or not certain experience should be required as a prerequisite for taking part in child and adolescent health state valuation. In contrast, however, we found that the majority of both UK and Swedish participants supported the idea that participants in health state valuation research should have experience of ill health themselves. This was also true for recommending prior experience caring for or working with children or adolescents, albeit with stronger support in the Swedish sample. It is thus interesting to juxtapose this against the approaches taken by NICE in the UK and the TLV in Sweden, with the latter appearing to be more aligned with public views than the former. This finding is likely to be of interest to NICE, given their policy on public involvement which stipulates that "lay people, and organisations representing their interests, have opportunities to contribute to developing NICE guidance, advice and quality standards, and support their implementation" [21].

As a subsidiary issue, participants were asked to rate several arguments proposed in the literature for and against the source and framing of values in child and adolescent health state valuation. Akin to previous qualitative work [22], the argument on who to ask that appealed most to the public was children's right to have a say, which is consistent with the United Nations Convention on the Rights of the Child (United Nations, 1989). The argument rated the least important was taxpayer perspective. A similar pattern of ratings has been observed in a sample of experts in child health valuation [30]. This is of note as this is the argument that has been used to justify methodological choices (of restricting valuation to adults) in the EQ-5D-Y-3L valuation protocol [16]. A number of studies are now beginning to converge that suggest that this argument is not received as convincingly as perhaps thought by a range of stakeholders, including members of the taxpaying public themselves [22].

The current study has investigated public views on the normative issues of who to ask and who to imagine when valuing child and adolescent health in principle. We are aware that this does not extend to the practical and methodological impacts of actually altering or implementing these normative positions. A number of studies have investigated the impacts of changing the source or perspective in child and adolescent health valuation with mixed findings [32-34] and this includes evidence that older adolescents can be meaningfully included as participants in valuation studies (e.g., [24]). If the normative positions of valuing

child and adolescent health are altered from those advocated, for example, in the EQ-5D-Y-3L protocol [16], then further methodological work will need to explore the most appropriate ways of implementing these changes. For example, some studies have advocated a preference for the use of discrete choice experiment (DCE) rather than time-trade-off (TTO) when adolescents are involved in valuation [29].

This study has several strengths, including a thorough collaborative development process for the supporting lay explanatory resources and survey itself; a dedicated and targeted sample of younger people (including 15-17 year olds); and a number of data quality checks to help maximise the quality of the data obtained. The study is also not without limitations. First, the sample was not designed to be representative of the wider population and oversampled adolescents. Thus, while providing an informative snapshot, findings may not generalise to the wider views of society. Second, while UK and Swedish participants were matched on a range of observables (including age, gender, education, and parental status), they were not matched on all sociodemographic indicators. Further analysis can be conducted to check the extent these differences affect responses and thus comparability of the two samples. Finally, while several layers of data quality checks were applied to help enhance the survey data, the limitations of online survey data apply, including potential inattentiveness and/or the potential for people to respond disingenuously. A number of indicators are available in the survey to denote the quality of responses (e.g., responses to video check questions, self-reported understanding, patterns of inconsistent responding) and subsequent work, including sensitivity analyses, will be conducted to assess the robustness of the conclusions from this study.

In conclusion, while some differences were observed in the strength of agreements, modal responses were similar across the UK and Swedish public on most normative issues. Most respondents thought that older children and adolescents should be involved in valuing child and adolescent health and that it would be acceptable for them to trade off life years. The majority of 15-17 year olds wanted to be eligible to value child and adolescent health. In context with other evidence, these findings can help to support normative methodological decisions made when valuing EQ-5D-Y measures and in the EQ-5D-Y-5L valuation protocol.

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