Exploring valuation of the adapted EQ-5D-Y-3L for 2-4 year olds: a think-aloud study

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Abstract

Objective: Valuation of health states for young children is necessary for including their health-related quality of life (HRQoL) in economic evaluations – but valuing HRQoL in young children presents challenges. This study aims to explore the views of the general public regarding 1) whose preferences should be sought 2) which perspective should be used 3) what age framing, 4) whether and how participants consider spillover effects, and 5) the feasibility of completing a discrete choice experiment (DCE) task for the adapted EQ-5D-Y-3L for 2-4 year olds.

Methods: Individual online semi-structured think-aloud interviews were used with Australian adults, including both parents and non-parents. The interview consisted of six think-aloud DCE tasks (imagining a hypothetical 3-year-old) using 12 different health states derived from the adapted EQ-5D-Y-3L for 2-4 year olds, followed by a range of open-ended questions to explore the key aims. Interview transcripts were double coded using an inductive and deductive approach and analysed using framework analysis.

Results: Interviews were conducted with 17 adult participants, including parents (n=9) and non-parents (n=8). Almost all participants found the task easy to understand and reported high confidence completing the task. Participants' views regarding whose preferences should be sought for the valuation of young children's health states varied widely, although there was a strong emphasis on parents or professionals who interact with children such as teachers or paediatricians. Despite the task framing asking participants to consider a hypothetical child, participants tended to instinctively think of themselves as a child, their own child, or a child they know. Several of the non-parents stated they would have responded differently if they were asked to think about a child they know, whereas parents stated that their responses to the task would be no different if they were asked to think about their own child or a hypothetical child. Participants thought about children who were either the 3-years or in an age range of 2-to-5 years. All participants stated that there would be no difference in their responses if considering a 2- or 4-year-old; however, almost all participants stated that they would respond differently if asked to consider a 10-year-old compared to a 3-year-old. Caregiver spillover effects were incorporated in valuation task considerations made by parents (e.g. how the child's health state impacts the parent's life), whereas future spillover effects (e.g. impacts on children's future health and non-health prospects) were considered by half the participants regardless of parental status. Almost all participants used information beyond the description of the task when making decisions, such as imagining if health states were treatable.

Conclusions: DCEs appear to be feasible for the valuation of the adapted EQ-5D-Y-3L for 2-4 year olds. There was strong consensus across respondents that the preferences of those with experience of young children may be the most appropriate source values. Framing the task for parents to consider a hypothetical child aged between 2-4-years old appears feasible. It is important for those conducting or using results from valuation studies to understand the types of considerations being made and the extent to which spillovers are being incorporated in participant's valuations of health states for young children. Our findings may have implications beyond the specific instrument used in this study (adapted EQ-5D-Y-3L) to valuation of health states in young children more broadly.

Introduction

There is a gap in the literature regarding paediatric health related quality of life (HRQoL) preference weighted measures for young children (i.e. under the age of 5). Currently there are an inadequate amount of research and guidance available to inform choices around valuation or application of value sets [1–4]. The EQ-5D-Y-3L is a concise, generic measure of HRQoL for children aged 5 to 18 years of age [5]. The EQ-5D-Y-3L is accompanied by value sets, enabling its use in estimating quality adjusted life years (QALYs)

for cost effectiveness analysis [6]. To fill the need for HRQoL measures in the young age range, a modified version of the EQ-5D-Y-3L was developed for use in children aged between two and four years [7]. Like the EQ-5D-Y, the adapted EQ-5D-Y-3L for 2-4 year olds consists of a descriptive system that comprises five dimensions of health: mobility, helping look after self, usual activities, pain and discomfort, and sad, worried, or unhappy. A three level (Y-3L) and five level (Y-5L) version of the adapted instrument have been psychometrically tested [8]. The adapted instrument was co-designed and developed as part of a qualitative study by Dalziel et al (2023) with HRQoL researchers, a paediatrician, a child psychologist, and parents of young children [7]. Parents of young children were asked to consider which aspects of the EQ-5D-Y-3L (in its current form) might be appropriate, as well as exploring what modifications of the instrument might be needed to improve the relevance and age-appropriateness for measurement of quality of life in young children aged 2-4 years [7]. Parents suggested that most of the existing domains were relevant, and minor changes were made to the wording to improve relevance to this young age range. Research has shown the adapted instrument to be reliable, age-appropriate, sensitive, and valid for the measurement of HRQoL in the young age range [8].

Value sets for HRQoL

HRQoL value sets provide preference weights for each possible health state described by a HRQoL instrument. For example, the EQ-5D-Y-3L has five domains that each have three response levels which results in a total of 243 possible health states (3^5) [9]. Value sets allow HRQoL health state descriptions to be converted onto a utility scale which is anchored at 0 and 1, zero typically being 'dead' and one being perfect health. These HRQoL values can then be used to calculate quality adjusted life years (QALYs) which are needed for economic evaluation of new interventions or treatments in a health technology assessment (HTA). HTA is necessary to provide evidence and guidance on societal decision making for interventions and technologies, with the aim to achieve allocative efficiency with limited resources. To create a value set, the stated preferences of the adult general population are typically sought. This is

both because adults are potential recipients of health care, and because they are taxpayers. It could be argued that the taxpayer should have a say in how health resources are allocated as they collectively fund and bear the societal healthcare costs [10]. There are multiple methods used to elicit individuals' preferences for health states, including composite time-trade off (cTTO), discrete choice experiments (DCE), or standard gamble (SG) [11]. DCE has been shown to be a valid and reliable methodology to elicit health state preferences of children, although it only provides the relative importance of dimensions and not values on the 0-1 scale [12,13]. This limitation of the DCE can be overcome through the introduction of duration as an attribute, or the use of some exogenous preferences information (e.g. relating to 'dead') to anchor [14].

Age-specific value sets for young child HRQoL

Given the adapted instrument has slightly changed the descriptors in health states (Supplementary Figure 1), it is unclear whether the value sets available for the EQ-5D-Y-3L for children aged 5-18 years, derived from adult's preferences considering views of a 10-year-old [5], are able to be used for the adapted EQ-5D-Y-3L for 2-4 year olds [15]. It is also unclear whether adults considering their views about health for an older child (i.e., 10-year-old) differ, or differ significantly, from their views about health for a younger child (i.e., 2-4-year-old). Although qualitative research suggest differences may exist, some quantitative studies have shown little to no impact on latent scale DCE and EQ-VAS values by varying the age framing, although there may not yet be substantial evidence to indicate otherwise [16–18]. If there are differences in values by age of child considered, a separate age-specific value set may be needed to accompany an age-specific HRQoL instrument, such as the adapted EQ-5D-Y-3L, before the instrument can be used for economic evaluation. Despite other HRQoL instruments being available for the measurement of QoL in the young age range (2-4 years old), such as PedsQL, CHU9D, or EQ-TIPS, currently only HUPS has a value set available (by linking to the value set for HUI3 [19]), thus limiting decision makers' ability to include young children in decision making for resource allocation [20–22].

Issues with developing age-specific value sets

Developing an age-specific value set for the adapted EQ-5D-Y-3L is problematic for a number of reasons. Most obviously, children under age 5 cannot be represented in the sample from which it is feasible to elicit preferences and some adults will have limited experience of children this age. Seeking adults' preferences for health states of young children raises issues of what perspectives they are asked to adopt [12,23]. Eliciting proxy-preferences through preference elicitation methods, such as DCE, for these young children generates normative and

methodological issues/questions, some around the perspectives sought and the framing of the task [24]. Currently, there is little to no guidance from HTA bodies, such as the Pharmaceutical Benefits Advisory Committee (PBAC) from Australia or the National Institute for Health and Care Excellence (NICE) from the United Kingdom (UK), regarding how we should approach valuation of children's HRQoL [25,26]. The HTA body from Netherlands released a new guideline in 2024 stating that EQ-5D-Y accompanied with Dutch value sets should be used for the paediatric HRQoL, and that for younger children a proxy version of the EQ-5D can be used [27]. Some of the issues and questions include: (1) Whose preferences should be used? For example, should the general population's preferences continue to be sought, or should an alternative approach be used such as using preferences from parents of children, child experts, or children themselves? (2) What should the perspective of the valuation task be? For example, the framing used may involve asking the adult what their preferences are while considering a young child (i.e., what they would prefer for a young child), or alternatively asking the adult what they think a young child's preference would be. Alongside this framing, the adult could be asked to take a perspective from a hypothetical child, a child they know, their own child, or themselves as a child (3) What age framing should be used? For example, for the adapted EQ-5D-Y-3L for 2-4 year olds this could be set as children aged 2-4 years, or a 3 year old as the midpoint. Finally, (4) what health or non-health considerations do adults think about when making their valuations? Do adults intentionally or unintentionally consider spillover effects (either impact on parents/carers or impact on the child's developmental goals, future education, work, and socio-economic prospects etc.) when valuing health states?

Aims

This study aims to explore the opinions of Australian adults who are non-parents and parents regarding (1) whose preferences should be used in valuing health states for 2-4 year olds (2) which perspective should be used (3) what age of child should be considered; and to assess (4) what factors and considerations adult respondents take into account when stating their preferences regarding health states in children 2-4 years old. In addition, this study aims to (5) explore the feasibility of using latent scale DCE to elicit preference for the adapted EQ-5D-Y-3L.

Methods

Study design

An exploratory qualitative study design involving a think-aloud DCE task followed by a semi-structured interview was used to explore parents' and non-parents' thoughts, choices, and views

on the valuation of young children's health states [28,29]. A phenomenological approach was taken, where it was assumed that there may be differences in thoughts between parents and non-parents due to the different experiences of/exposure to young children (i.e., assuming parents would have more experience and exposure to young children compared to non-parents) [30].

This study was granted ethical approval by the University of Melbourne Human Research Ethics Committee, Melbourne, Australia (Number: 2023-26964-43277-3). The findings of the present study were cross-checked and aligned with the consolidated criteria for reporting qualitative research (COREQ) checklist [31].

This work builds on an existing think-aloud study aimed at understanding valuation of health states for older children [32] as part of the QUOKKA research programme [33].

Sample

Participants were recruited during August 2023, with all interviews conducted and completed in September 2023. Participants were recruited through a market research agency (Focus People) located in Australia. The market research agency sent out emails through their pre-existing, participant database to identify and recruit eligible participants across Australia. Participants were screened for eligibility and were provided with a study information form before providing informed consent to participate in the study. Eligibility included: over 18-years-old, residing in Australia, English speaking, and access to a laptop with a working camera and microphone. Purposive sampling was used to get an even distribution of parents and non-parents, as well as a wide range of ages and an even spread of gender [34]. Of those that were selected to participate in the study none opted out or refused participation.

Reflexivity

All interviews were conducted by AVH, a Male PhD candidate at the University of Melbourne (UoM), Australia and a non-parent. AVH has experience in conducting interviews with participants, and has been formally trained in interviewing and qualitative research. Interviews were double coded by AvH and RJ. RJ is a non-parent Female PhD candidate at the UoM and has formal training in qualitative research. The analytical framework was finalised with the broader research team. No researchers in this study were known to any of the interviewees.

Data collection

Interviews were conducted online via a video conferencing platform Zoom [35]. The interview involved a think-aloud methodology to attempt to gain insights into the cognitive processes involved in decision making. The interview consisted of eight stages (Supplementary Figure 2): 1) introduction to the study, 2) explanation of quality of life, 3) self-completion of EQ-5D-

3L and EQ VAS, 4) think-aloud explanation and practice, 5) DCE think-aloud tasks (using think-aloud methodology), 6) semi-structured interview, 7) demographic information, 8) difficulty and experience questions. The interview structure was pilot tested with three of the authors (RJ, KD, and ND) before conducting interviews with the participants. After a brief explanation, participants were asked to report their own health-related quality of life using the EQ-5D-3L and EQ-VAS. The EQ-5D-3L is an adult quality of life measure that contains five dimensions of health (mobility, looking after self, usual activities, pain and discomfort, anxiety and depression) with three levels of severity (no issues, some issues, or a lot of issues) [36]. EQ VAS is a visual analogue scale that ranges from 100, best imaginable health, to 0, worst imaginable health [36]. The participants were then given an explanation of the think-aloud task and were provided with an unrelated example think-aloud DCE task asking the participant to choose between two different travel destinations with five different attributes. A DCE is a pair-wise choice task which involves participants choosing between either option A or option B, where each option has the same set of attributes with differing levels. A think-aloud method involves asking participants to verbalise and articulate their thought processes as they completed the task [28,29]. The interviewer would remind the participant to continue verbalising aloud their thought process if the participant was silent for too long, and would ask open-ended questions (e.g., "why did you choose A instead of B?") if the participant gave an answer without providing their thought process. The participants then completed a set of six DCE tasks (12 different adapted EQ-5D-Y-3L health states) while considering or thinking about a hypothetical 3-year-old and thinking aloud. In each case, the participants are asked to select the health state that they prefer, or think is better. In this case, the DCE attributes reflect the five domains of the adapted EQ-5D-Y-3L. The health states selected for the DCE task were chosen from a similar think-aloud study using the EQ-5D-Y-3L [18] (Supplementary Figure 2), and were expected to engage and stimulate the participants' thought processes due to the variation in dimensions and levels. An example of the task is found in supplementary material (Supplementary Figure 3). The order of presentation of the set of DCE tasks was randomised for each participant. Following the think-aloud task the participants completed a semi-structured interview where an interview guide was used to ensure all key questions were covered and to provide consistency across all interviews. At the end of the interview, participants completed basic demographic questions and participants provided feedback on the difficulty of the tasks. Transcripts were not returned to participants for commentary or correction, and participants were not asked to provide feedback on the overall study findings.

Data analysis

A seven stage framework analysis was used to analyse the interview data [37]. Framework analysis was chosen as it offers analysis of the qualitative data in a systematic and structured approach, while providing flexibility in the approach taken to develop the themes. The seven stages process follows: 1) transcription of the data, 2) familiarisation of the transcript, 3) coding of the transcript, 4) development of an analytical framework, 5) applying the analytical framework to the transcripts, 6) charting into the framework, and 7) interpretation of the data. Stage 1-2: The audio recordings were transcribed and the two coders (AvH and RJ) familiarised themselves with the transcripts. Stage 3-4: An iterative process was used to develop the analytical framework where each coder coded three transcripts independently per round. After each round the coders compared themes identified and discussed the overall analytical framework. The *a priori* position of the study was to align data analysis with the preconceived aims, therefore, both a deductive (i.e., applying key aims to define themes) and an inductive approach (i.e., allowing themes to appear naturally) was used to develop the analytical framework. The coding process reached saturation after three rounds (i.e. , no new themes identified). Stage 5: The analytical framework (Table 1) was then discussed and agreed upon with the wider research team (triangulation of information) before being applied to all transcripts. The analytical framework codebook contained seven higher themes, five through a deductive approach (Whose preferences, Perspective framing, Age framing, Spillover effects, and Feasibility of task) and two through an inductive approach (Approach for deciding preferences, and How respondents viewed 3-year-olds) (Table 1). Stage 6-7: The data were then charted and interpreted with a phenomenological theoretical approach [30]. NVivo 14.0 software was used for the framework analysis [38].

Table 1: Analytical Framework Codebook

	Themes	Sub-themes
1) Approach for deciding preferences	a) Focusing on what's important for a young child	
	b) Strategy deciding between A and B	i) Reduce number of states to consider ii) Overall severity iii) Linking to medical condition iv) Differing number of issues v) Which states could be treated or supported
	c) Importance of levels	i) Some issues throughout is better than few issues that are severe ii) No issues and some issues are the same
	d) Domain connectivity	i) Sad, worried, and unhappy linked to pain and discomfort ii) Conceptualise child in each health state by connecting domains
2) Whose preferences*	a) General adult population	
	b) Caregivers and parents of children	
	c) Non-parents	
	d) Children	
	e) Professionals	
	f) Lived experience	
3) Perspective framing*	a) Framing difficulties	
	b) Difference in framing	i) Comparing own child vs hypothetical ii) Comparing child you know vs hypothetical
	c) Framing method used	i) Thinking of someone they know ii) Thinking of own child iii) Thinking of self as a hypothetical parent with a child iv) Thinking of self
4) Age framing*	a) Who they thought about	
	b) Preferred age-framing	
	c) Differing views on age framing	i) Young child (under 5) ii) Older child (over 5) iii) Adult age
5) Spillover effects*	a) Future spillover effects	i) Health will naturally improve over time ii) Lifelong impact of health iii) Impact on development iv) Life experiences considered
	b) Caregiver spillover effects	i) Burden on caregiver ii) Financial cost iii) Emotional burden iv) Caregivers can support or attenuate quality of life concerns
	c) Beyond the description of the task or health state provided	i) Health can be improved ii) Health resources and treatments are available iii) Child naturally has health issues
6) Feasibility of task*	a) Experience completing DCE	i) Difficulties ii) Emotional
7) How respondents viewed 3-year-olds	a) Ability to communicate	
	b) In the middle of independent and dependant	
	c) Not sociable	
	d) Hard to tell what is normal or an issue at this age	
	e) Health state less worse in 3-year old than older children or adult	
	f) Normal for children to have issues at this age	
	g) Mental health hard to spot	
	h) Differing views for a young child's health state	i) Child too young to experience anxiety or depression ii) Degree child feels pain not the same as adult iii) Child doesn't know any better

*Themes and sub-themes developed through inductive approach, unless specified. * Represent deductive themes.*

Results

A total of 17 participants, 9 parents and 8 non-parents, completed the think-aloud interview. There was a relatively even split of gender and educational status, as well as a wide distribution of ages among both groups (Table 2). Half of the parents had at least one child under the age of 4, with the rest having children over the age of 5 (Table 2). On average the interview time was 54 minutes.

Table 2 Demographic information on the sample of Australian adults participating in the think-aloud DCE valuation study of adapted EQ-5D-Y-3L for 2-4 year olds.

	Parental Status (N (%))	
	Parent (9)	Non-parent (8)
Gender		
Male	5 (56%)	3 (38%)
Female	4 (44%)	5 (62%)
Age Group		
20-29	2 (22%)	2 (25%)
30-39	3 (33%)	2 (25%)
40-49	2 (22%)	2 (25%)
50-59	2 (22%)	2 (25%)
Education*		
No qualification	0 (0%)	0 (0%)
Completed year 12 and/or Cert III	3 (33%)	2 (25%)
Diploma or Advanced Certificate	4 (44%)	2 (25%)
Bachelor's Degree or Higher	2 (22%)	4 (50%)
Income*		
<$26,000p/y	0 (0%)	0 (0%)
$26,000-$65,000p/y	3 (33%)	2 (25%)
>$65,000p/y	6 (67%)	5 (63%)
Prefer not to say	0 (0%)	1 (13%)
Employment status*		
Full time	5 (56%)	5 (63%)
Part-time	0 (0%)	1 (13%)
Self-employed	0 (0%)	1 (13%)
Full time carer	3 (33%)	1 (13%)
Unemployed	1 (11%)	0 (0%)
Marital Status *		
Single	3 (33%)	3 (38%)
Domestic partnership	0 (0%)	1 (13%)
Married	5 (56%)	4 (50%)
Divorced	1 (11%)	0 (0%)
State		
VIC	3 (33%)	2 (25%)
NSW	2 (22%)	1 (13%)
QLD	1 (11%)	1 (13%)
SA	2 (22%)	1 (13%)
TAS	0 (0%)	1 (13%)
ACT	0 (0%)	2 (25%)
WA	1 (11%)	0 (0%)
Work experience with children		
Yes	1 (11%)	3 (38%)
No	8 (89%)	5 (62%)
Number of own children		
1	5 (56%)	-
2	4 (44%)	-
Own children's age		
<4	5 (56%)	-
>5	4 (44%)	-

** Variables derived and modified from Australian Bureau of Statistics [39]*

Approach for deciding preferences

For all DCE tasks there was an observable similar patterns in the responses by both parents and non-parents in their preferences across all six choice sets (Table 3). While making decisions between which health state was considered better, participants tried to focus on what was important and what their preferences would be when thinking of a hypothetical 3-year-old child. A wide range of strategies were used to assist with decision making such as: reducing the total number of domains to consider (i.e., completing ignoring domains that were the same between A and B), assessing overall severity, comparing the differing number of issues between A and B, and considering which health state could be more easily treated or supported. When assessing overall severity of the health states, some participants noted the importance of levels indicating some level 2 issues across multiple domains was better than having a few issues at level 3. This could describe why all participants chose B for the choice set (A) 11332 vs (B) 22222 (Table 3). A few participants linked domains together which may have increased or reduced the participants perception of the overall severity of the health state ["*pain can be managed to a certain degree but if they're not worried, sad or unhappy they would be in a better place to deal with that pain*" ID 6 – Parent]. The majority of participants had a clear preference of a particular domain/s which were given a greater weighting than others when making the decision. Which domain was given more weight varied with some participants prioritising physical domains (i.e., usual activities, helping look after themselves, mobility), and a majority prioritising the sad, worried, or unhappy and the pain or discomfort domains ["*I was thinking more from the point of view of the bottom two rows being the most important. So whether they're sad, happy or worried, and whether they are in pain or discomfort*" ID 14 – Parent], with a few participants swapping the domains they considered most important in between the tasks.

Table 3 DCE responses of parents and non-parents for six adapted EQ-5D-Y-3L health state sets

		Adapted EQ-5D-Y-3L health state sets					
ID	Parental status	11332 (A) vs. 22222 (B)	13213 (A) vs. 32331 (B)	11113 (A) vs. 11121 (B)	31231 (A) vs. 32313 (B)	33323 (A) vs. 21133 (B)	33311 (A) vs. 11133 (B)
1	Yes	B	A	B	A	A	A
2	No	B	B	B	A	B	A
3	Yes	B	A	B	A	B	B
4	Yes	B	A	A	B	B	A
5	No	B	A	B	A	B	B
6	Yes	B	B	B	A	B	B
7	No	B	A	A	A	B	A
8	No	B	B	B	A	A	A
9	No	B	A	B	B	A	A
10	Yes	B	A	B	A	B	B
11	No	B	A	B	A	A	A
12	Yes	B	A	B	A	A	A
13	Yes	B	A	A	A	B	B
14	Yes	B	B	B	A	A	A
15	No	B	B	B	A	B	B
16	Yes	B	A	B	A	B	A
17	No	B	A	B	A	B	B
A	Yes	0 (0%)	7 (78%)	2 (22%)	8 (89%)	3 (33%)	5 (56%)
B		9 (100%)	2 (22%)	7 (78%)	1 (11%)	6 (67%)	4 (44%)
A	No	0 (0%)	5 (63%)	1 (13%)	7 (88%)	3 (38%)	5 (63%)
B		8 (100%)	3 (38%)	7 (88%)	1 (13%)	5 (63%)	3 (38%)

Whose preferences

There were a wide range of thoughts on whose preferences should be sought for the valuation of young children's health states. The differing thoughts were reflected in the various themes. Themes include: general adult population, caregivers of children, adults and children's perspectives, those with experience with children including professionals, non-parents, and those with lived experience of health (Table 4).

Table 4: Illustrative quotes regarding whose preferences should be sought for the valuation of the adapted EQ-5D-Y-3L for 2-4 year olds

Themes	Illustrative quotes*
General adult population	*"It would be great to do it [DCE valuation task] across a wide demographic, the socioeconomic demographics as well. Because I feel it will have varied outcomes dependent on those environments"* **ID 4 [Parent]** *"I always think it's good to get a broad range, for accuracy"* **ID 7**
Caregivers and parents of children	*Probably just parents, I would say, because people without children don't really have any idea about kids. Of course I think there needs to be professionals involved and if they're non-parents, then that's fine, but I don't think asking the general population, like non-parents about a child's health is, yeah, no, I don't think they should be involved […]. You don't really know what it's like to have a child or until you have one. You think you do, but you don't."* **ID 13 [Parent]** *"At that age, I think probably the parents are the ones who would know better about what ... what's best for their child […]. I think you need to have that close experience with children to be able to ...accurately ...give that priority ranking."* **ID 14 [Parent]**
Adults and children	*"I think it's important to get both perspectives of a child and an adult […] I don't feel like you could ask a three year old what they prefer because I feel that they wouldn't know"* **ID 5** *"It would be good to get the children to do it, but again, I don't know how you'd be able to do it. The questions for me, even I had trouble trying to compare things […] it would be good to see what they think because they've got their own thoughts."* **ID 15**
Experience with children & professionals	*"I also think say school teachers and childcare workers and people that deal directly with young kids. […] I've got nieces and nephews, and I actually care about, even though I don't have my own human kids, I still have a connection with my nieces and nephews, and take an interest in what's what."* **ID 17** *"Child psychologists or […] a paediatrician, child doctor, they would probably the best ground runners to determine then where and what was needed from here on."* **ID 7**
Non-parents	*"A perspective of just common person who have seen a few things around and don't relate it to somebody you know or your own, just as general thinking. So I would go with the same approach because if I go to parents, then based on their circumstances, their responses would vary quite a lot."* **ID 9**
Lived experience of health	*"opinion of adults who had been through those situations as a younger child"* **ID 3 [Parent]**

**To enhance readability of the quotes non-essential information within quotes has been replaced with ellipses […].*

There were some thoughts around ensuring a representative sample of the general population to get a wide range of perspectives and because it would be fair as everyone would get their chance to "have their say". There was a strong emphasis on seeking parents' perspectives, although this was mostly suggested by parents, for the reasons of "they know more", are more experienced, and are more accurate. On a similar note, there were a handful of reasons to not include non-parents, as non-parents have less experience, and may not have an interest or understanding of young children's lives. There were some opposing views on seeking parents' preferences expressing that parents are too biased in their decision making. There was also a strong preference towards those with experience of children whether professional (e.g., paediatrician, child psychologist, childcare worker, developmental researcher, educator) or personal (e.g., having nieces or nephews and friends children). Participants expressed that professional expert knowledge or experience with children was beneficial to know what is needed for them. Participants also liked the idea of including children, however, there were concerns that children in this age range (2-4 years) are too young to complete such a cognitively challenging task.

Perspective framing

Framing difficulties

There was wide variability with participants either finding the task of thinking of a hypothetical 3-year old easy or challenging. Those that suggested the framing of the task was easy referred to the fact that they have experience as a parent, whereas those that found the task challenging commented on how they either did not have any experience with children (or children with health conditions), or that they found the task to be emotionally challenging.

Difference in framing

Some of the non-parents stated they would have responded differently to the task if they were asked to think about a child they know, whereas all of the parents stated that their responses would be no different if they were asked to think about their own child or a hypothetical child.

Framing method used

Despite framing participants to consider a hypothetical child, participants tended to think of their own child, or a child they know. In almost all cases, parents thought of their own child/ren at some point during the task. Parents that had healthy children used their child as a reference point to think about a hypothetical 3-year old, compared to parents that had children with existing health conditions who thought of their child instead of a hypothetical child. Similarly, some of the non-parents thought of a child that they know to help with setting a reference point.

Age Framing

Who they thought about

The age participants actually thought about were either the 3-years (as stated in the task) or an age range of about 2-to-5 years. Participants referred to this age range as toddler or pre-school age, and viewed the child at an age where they were not fully dependant like a baby ["*not total baby but three being potentially they can walk, they can talk*" ID 5], but not fully independent like a school child ["*[child] definitely can't, is nowhere near being able to look after themself so is definitely very, very reliant on a parent or guardian in that sort of thing*" ID 1 – Parent]. Some participants commented that they would have preferred to have thought of an age range rather than a particular age ["*I couldn't understand why it was just targeting that one age .. and why not say between 0 and 5, or 3-5. Why just the 3-year-old.*" ID 7]

Differing views on age framing

Almost all participants suggested that their responses to the valuation task wouldn't have changed when considering a child aged 2 or 4 ["*Two-year-old or three-year-old, I don't know if I would've thought any differently*" ID 6 – Parent]. Conversely, most participants said that their responses to the valuation task would be very different if thinking of a much older child

(i.e., child aged 10 or older). Some reasons participants provided explanations for why they might respond differently such as putting more emphasis on the physical domains (i.e., mobility, looking after self, and usual activities) ["*I'd probably start to be putting a bit more importance on mobility issues and those types of things"* ID 14 - Parent] and the fact that older children have more capacity to manage or deal with pain or mental difficulties ["*I feel like a 10-year-old can more than likely handle pain a little bit easier or ...can just handle pain a little bit more than a two, three, four-year-old"* ID 13 - Parent].

Spillover effects

Future spillover effects

Future spillover effects were observed for several participants regardless of parental status (Table 6). Some participants expected that certain decrements within the health state would naturally improve over time, thus appearing to put less weighting for those domains. A few participants had concerns that certain domains could result in worsening health or complications in the long term putting more weight for those domains. For example, having 'a lot of pain or discomfort' needing long term pain medication use could lead to drug addictions in the future. Given the child's young age, many considered how the different health states might impact the child's developmental progression or needs as they aged. In addition, some participants considered how the different health states for the young child might impact future life experiences such as education, social connectivity, and employment or financial prospects.

Caregiver spillover effects

Caregiver spillover effects were mostly observed for parents, except for few instances where non-parents put themselves in the shoes of a parent and considered how the hypothetical child's health state might impact them. Most parents considered the overall impact or burden of the child's health state on the caregivers in a broad sense, such as the level of involvement required from the parent. Whereas some considered specific impacts to the caregivers life such as the financial and or emotional cost of the different health states on the hypothetical caregiver. Some participants considered the degree in which parents can support or attenuate aspects of the child's health, including the 'helping look after self', 'usual activities', and 'mobility' domains.

Beyond description of the task or health state provided

The majority of participants pulled in information beyond the description or health state provided. Several participants considered that certain aspects of health can be improved, and others considered what healthcare resources or treatments might be available or accessible, or not available or accessible, to help optimise or manage aspects of the young child's health state.

Finally, some participants had particular views on a 3-year-old child and contemplated what decrements in the health state would be normal for a child at this age.

Table 5: Illustrative quotes regarding the type of spillovers effects found when completing a latent scale DCE valuation of the adapted EQ-5D-Y-3L for 2-4 year olds

Themes	Sub-themes	Illustrative quotes*
Future spillover effects	Health will naturally improve over time	*"a lot of problems with movement, that's something that can I think be solved fairly easily; they'll probably be able to walk, run and jump soon."* **ID 8**
	Lifelong impact of health	*"But I suppose with the socialising one, or with the running or anything like that, it felt like that also included in the future, where I suppose it would affect them daily"* **ID 2** *"You are talking medication that could be life long and develop into an addiction when they are older […] if it's really strong medication."* **ID 16 – Parent**
	Impact on development	*"I was probably thinking today ...and how that will impact their developmental health state in the future."* **ID 4** *"I suppose longer term impacts...on how the child is going to develop or how the child is going to progress throughout their life […] If it's not addressed at that early stage and they're unhappy and sad and not getting any joy from life that could manifest into some real significant issues ... when they're older."* **ID 16 – Parent**
	Life experiences considered	*"The problems with mobility, it sounds to me like would they be wheelchair-bound or something along those lines, if they're having problems now, what can they accomplish in life? […] normal things like graduating, […] going to school, […] travelling, there's always going to be boundaries"* **ID 1 – Parent** *"Being able to get a job, to get money, to be able to afford cost of living. […] Would they be able to survive and look after themselves without family around them"* **ID 17**
Caregiver spillover effects	Burden on caregiver	*"A lot of problems with usual activities. I think that's quite a big difficulty in a child's life and for the parents."* **ID 10 – Parent** *"I did think about parents as well [...] for example, if your child can't walk and they're in a wheelchair, then that's a huge thing for you as a parent."* **ID 13 – Parent** *"I was thinking of what would I be looking for or prefer if I was the parent of that child […] because obviously it would have a huge impact on the parents"* **ID 5**
	Financial cost	*"Finances, because it will cost money for whatever we need to do. I mean, mental health, physical health, everything as an adult, it costs so much money."* **ID 15**
	Emotional burden	*"I thought about the parents because I've seen firsthand a colleague […] and the trouble that she went through, the pain she went through. It was very tough. […] I thought about that, those impacts, like what parents go through and what the support family go through. Everybody's involved, of course. […] But parents are the one who are impacted the most, both financially, emotionally, physically"* **ID 9**
	Caregivers can support or attenuate quality of life concerns	*"Having lots of problems with movement […], they don't know any better. A lot of problems with helping look after themselves, you're there, you'll help them. And the same with the usual activities, if they can't do something and you're there to help them, they don't know any better."* **ID 6 – Parent** *"Usual activities, you have a lot of problems. It is not ideal, but you might get some help around it. Helping with washing. This, again, not ideal, but you can get some help around it. Walking, running, jumping, you can get some help around it."* **ID 9**
Beyond the description of the task or health state provided	Health can be improved	*"They've got a lot of problems helping look after themselves, and that's something that can be taught."* **ID 10 – Parent**
	Health resources and treatments are available	*"With pain and discomfort we could go down the route of with a doctor and medication and we can alleviate that"* **ID 6 – Parent** *"If there was a deficit in one area straight away, my mind would think, okay, but we can go and see an OT or we can go and see a physio and they can help with this issue. ."* **ID 10 – Parent** *"So for me, the key factor only there is their emotive state, which is worried or sad or unhappy, which I think, for a three year old, can be altered in a positive environment."* **ID 4 – Parent**
	Child has some issues as part of natural development	*"A three year old is not expected to be able to be fully confident with walking, running and jumping […]. It is also normal for them to have problems helping look after themselves, like washing themselves, dressing, and going to the toilet by themselves; that's normal."* **ID 8**

**To enhance readability of the quotes non-essential information within quotes has been replaced with ellipses […].*

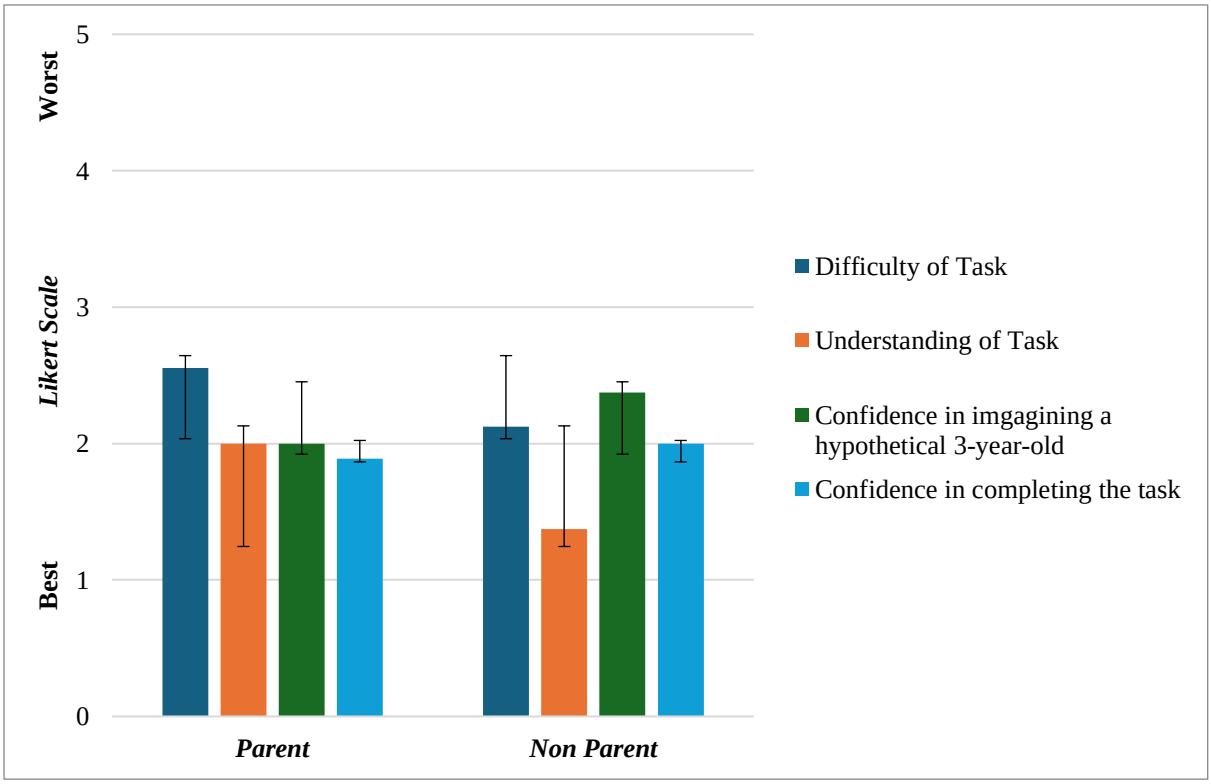
Feasibility of the task

Experience completing the DCE

There were a wide range of thoughts and feelings around the difficulty on completing these valuation tasks when thinking of a hypothetical 3-year-old. Some found the task emotionally challenging ("depressing", "sad", and "awful") having to consider such a young child living in such a poor health state, particularly when both health states being considered were equally severe. On the other hand, some non-parents found the task challenging, and somewhat "stressful", due to not having any experience or knowledge of young children to draw upon or relate to while making these decisions. Other participants did not find the task challenging or difficult, and approached the task "*diplomatically and trying not to get emotionally involved*" (Pt 3 – Parent).

Both parents and non-parents appeared to have a good understanding of the task, found the valuation task somewhat easy to complete, were quite confident in completing the task, and were quite confident in taking the perspective of a 3-year old (Figure 1).

Figure 1 Participants' feedback on their experience completing the DCE valuation tasks for states described using the adapted EQ-5D-Y-3L for 2-4 year olds.



A five point Likert scale ranging from 1 (best) to 5 (worst); 1 being most confident/understood (least difficult) and 5 being least confident/understood (most difficult). Average responses displayed. Mean score with standard deviation displayed.

How respondents viewed 3 year olds

Participants had a wide range of views of 3 year olds. Some participants viewed 3 years of age as being too young to know and verbalise their mental state, with some thinking children at this age are too young to experience such emotions ["*Just the thought of thinking a three year old would be anxious or depressed, I think you're too young to even know what those feelings are and experience them*" ID5]. Other thoughts include children this age as being in this intermediate state of being dependant and independent simultaneously, and that it is normal for 3 year olds to be in certain health states. Some participants commented that children at this age don't necessarily know any better, thereby minimising the weight of certain health domains (particularly the physical health domains of mobility, helping looking after self, and usual activities). When assessing 'some' or 'a lot' of pain, some participants noted that the degree to which younger children feel pain is more than in older children or adults due to having a lower tolerance or threshold to pain.

Discussion

This study is the first to qualitatively explore the valuation of a EuroQol measure for very young children and provides evidence to support the development of value sets for instruments in this age range. Overall, all of the participants, regardless of parental status, found the latent scale DCE using adapted EQ-5D-Y-3L health states easy to complete, and expressed high confidence in completing the task while considering a hypothetical 3-year-old child. In the small sample, it was observed that parents and non-parents had similar patterns in their DCE choices, with most participants placing a higher weighting on mental and emotional aspects of HRQoL.

The qualitative analysis indicated that almost all of the participants preferences towards the DCE tasks wouldn't be any different when considering a 2 or 4 year old compared to a 3 year old. However, the majority of the participants noted that their choice would likely differ if asked to consider an older child, aged 10 years old, compared to a 3 year old. These findings are comparable to other qualitative studies, such as Reckers-Droog V et al (2022), that adults state that they would have different preferences when considering different age children [18]. These differences however don't necessarily play out when explored quantitatively. One theory for the paradox of preferences could be that an individual's preferences between different age groups are only different for very few health states or domains, which may result in marginal differences in health state ordering. Alternatively, the differences may simply 'wash out' when modelled at the overall level based on valuation data from large samples.

Assuming there are very minimal differences in health state values for states pertaining toa young child and an older child framing then a pragmatic approach could in principle be adopted whereby the value set for older children could be used for a young child instrument (i.e., application of the value sets which are available for the EQ-5D-Y-3L to the adapted EQ-5D-Y-3L). Conversely, assuming there are significant differences in preferences between different paediatric ages and age-specific value-sets are considered necessary, future valuation studies asking participants to consider a hypothetical child aged between 2-4 years old appears appropriate and feasible.

Whose preferences should be used

There were a wide range of thoughts from the interviewees on whose preference should be sought to generate an age-specific value set for the adapted EQ-5D-Y-3L for 2-4 year olds. There was general consensus towards parents or those with experience of young children being most appropriate to complete the valuation tasks. Nevertheless, whose preferences are relevant is a normative question and to date the current approach towards developing preference weights for adults has been to use the general adult population with a 'taxpayer argument' (i.e., those that are of working age collectively contribute and bear the burden of the healthcare costs in the society, therefore, it is the belief that their thoughts or preferences should be used). Whilst this may be appropriate for adult valuation, this may not necessarily be the case when considering a children's health states. A systematic review by Bailey et al (2022) highlighted the variability of whose preferences are currently used for paediatric valuation of health states, with most studies using adults and some using a parent, healthcare provider, or the child/adolescent themselves [11]. However, when reviewing the few valuation studies that have included health states for children under the age of 5; two studies used preferences of half parents and half general adult population [40,41], two had a mix of health professionals and parents [19,42], with one having only parents [43], and one with only young adults (university students) [17].

Despite participants being asked to think of a hypothetical child, most participants thought about a child that they know of (or their own child) while completing the valuation tasks. Interestingly, when asked if there would be any differences in their preferences if they were to think of a child they know (or their own child) versus a hypothetical child, the majority of the parents stated there would be no differences in their preferences, whereas several non-parents suggested that they would think differently. These qualitative results suggests that potentially parents/caregivers approach might be more consistent in terms of the framing of the child in

the valuation task, and simply asking respondents to think of 'a child' may be more appropriate than a 'hypothetical child' given participants think of a child anyway.

Although this study was conducted in an Australian context, the above approaches on whose preferences should be sought for paediatric value sets is applicable to other HTA bodies around the globe. We propose four thoughts for HTA bodies to consider. Firstly use of a general population would maintain consistency for both adult and paediatric valuation studies. One important aspect to consider is whether the entire general adult population is capable of imagining the preferences of a young child, to ensure adequate knowledge to make informed judgements. Further, this argument on knowledge and experience of what is being valued applies to any value set development. Secondly, parents and/or those with experience with young children greater knowledge and understanding of what a young child's life looks like, but also may come with their own biases. Thirdly, collecting preferences from both a general adult population and a parent sample allowing users to select and choose the most appropriate responses, whether choosing just the parent sample or choosing a combined sample. This would allow users of the value set to transparently see differences between the two samples. A key consideration is whether there are any differences in preferences for young children between non-parents/those without experience and parents/those with experience of young children. If there are no differences, then it may not be necessary to take an alternative approach to the most commonly used general population. However, if a difference does exist, then a normative decision needs to be made . Additional research is needed to explore these differences, although defining what counts as 'experience with children' will be challenging. A fourth option of children valuing their own health states is considered unfeasible for 2 to 4 year olds due to lack of understanding. Using older children to complete valuation tasks for younger children is possible, although this would pertain similar issues to adults valuing a child (i.e., valuation of someone else). Few participants in the study mentioned that obtaining preferences from children in addition to adults would be idea, however, there were concerns over feasibility. A recent roundtable discussion in the United States (US) by Nazari et al (2022) indicated that key stakeholders (including paediatric clinicians and academics, HTA bodies, health economic and outcomes research professionals, and lay persons) felt that adolescents would be able to relate to a 10 year old child health state more so than adults and were likely capable of self-completing valuation tasks [44]. It is possible that adolescents could also relate to a 2-4 year old health state. Further, the stakeholders in the Nazari et al (2022) study had concerns that adults would be inconsistent in their views about a child, though some suggested parents may be suitable [44]. A similar roundtable discussion in the UK by Powell (2024) showed positive support for

the inclusion of children that are of a mature or understanding age [45]. Further, the roundtable discussion showed strong support for seeking parents perspectives, particularly for valuation tasks of young children health states [34].

Management of spillover effect

Spillover effects in valuation tasks are distinct from spillover effects in the measurement of HRQoL. Spillover effects in valuation tasks of health states could cause inconsistencies and lead to a bias upward or downward on the utility values generated for certain health states where such spillover effects are or are not considered. As such, it is important for those conducting or using valuation studies to understand the types of considerations being made and the extent to which spillovers are being incorporated in participant's valuation of health states for young children. The results of this study suggest caregiver spillover effects are mostly relevant when looking at a sub-sample of parents completing the valuation task. This is intuitive as parents have been or are currently caregivers of children, therefore, they have likely have an understanding and experience in how a child's health state directly impacts themselves as caregivers. As such, it is possible that differences that may be observed between parents and non-parents in valuation tasks could be attributable to either differences in experience of children and/or caregiver spillover effects in the parental group.

There are three possible ways to deal with spillover effects in future valuation studies. First, the use of guidance notes and improved framing techniques to minimise spillover effects as much as possible, though it may not be possible to fully prevent. For example, reminding the participants in each task to not consider how the health state may impact caregivers or asking participants to assume there are no possible treatment options available for the health states being considered. A second approach, aiming to measure and assess the value or magnitude of the spillover effect/s, which could then be accounted for when developing utilities, or measuring QALYs for cost-utility analyses. A recent systematic review by Lamsal et al (2023) summarised the current analytical approaches taken in integrating caregiver/family spillovers for paediatric cost-utility analyses, with one method that could be used for utilities (1) incorporating family spillover effect using a multiplier approach [46], and (2) applying the disutility or utility decrements of the child's illness on the caregiver onto the child's utility [47]. For example, for a health state that has mobility at 'a lot of problems', caregiver spillover effect might cause an increase of 0.08 on the utility scale, therefore, subtracting 0.08 from the overall utility score in these health states would be necessary if the aim is to get isolated utility scores unimpacted by caregiver spillover effects (disutility of illness for caregivers). In the above example caregiver spillover effect might cause an increase in the utility score (i.e., be of

better health) due to the caregiver being able to attenuate 'a lot of problems' with mobility, alternatively may cause a decrease in the utility score (i.e., be of worsening health) as the health state is causing an emotional or physical burden on the caregiver. Measuring the size of caregiver spillover effect and how it varies for different health states could have wider implications to other paediatric HRQoL valuation studies. There are only a few studies in the literature that have measured the disutility of caregiver spillover effects, with values ranging from no effect to as large as -0.718 [48]. Third, a combination of the first two methods to deal with spillovers. For example, task framing could be used to minimise future and consideration beyond the task spillover effects, while caregiver spillovers could be dealt with quantitatively. Accounting for caregiver/family spillover effects, both costs and effectiveness (i.e., utility score or the QALY), appears important given consideration of the spillovers trends to result in a more favourable CUA result for paediatric interventions, where the incremental-cost effective ratio (ICER) moves below a common cost-effective threshold [49]. Consensus is needed on the most appropriate method/s used to account for these spillovers is needed.

Limitations

Comments from a few participants suggested that the domains of the instrument did not necessarily apply well to a 3-year-old child. This is because these few participants felt that children at this age have difficulties or issues that would be considered normal for the particular child's age, such as helping to look after themselves or being fully mobile. This could be due to the instrument not performing optimally and picking up problems that are not really there, or it could be due to the layout of the DCE task to reflect the guidance notes that were provided to help participants think through an age appropriate interpretation. The 'helping look after themselves' was a particularly difficult and confusing domain to understand for a few participants, although mostly for non-parents. Further developing lay summaries for what is meant by the dimensions in the context of a 2-4 year old would be useful to support the valuation process.

The DCE approach used in this study did not include any anchoring methods, such as duration or death, and cannot be used to generate value sets. Anchoring in this age group is a key area that needs to be resolved if value sets are required in this group, and should be the subject of future theoretical and empirical work. Alternatively, the latent scale DCE could be anchored with cTTO tasks, as is the case with the EQ-5D-Y-3L[5].

Conclusion

Latent scale DCE appears to be feasible for the valuation of the adapted EQ-5D-Y-3L, although alone would not be able to generate values anchored at 0-1 as required for QALY estimation. As such, further exploration of alternative means of anchoring these values is required. There was strong consensus across respondents that valuation tasks are most appropriately undertaken by those with experience of young children. Those using the information may however prefer to separately see and compare preferences of those with experience of children alongside a general population sample more traditionally used to generate value sets. Framing the task for participants to consider a child age range between 2-4-years old appears preferred and feasible with participants likely to think of a child they know regardless of how instructed. It is important for those conducting or using results from valuation studies to understand that participants completing young child health state valuation are likely to also consider caregiver burden and future outcomes for children. Our findings may have implications beyond the specific instrument used in this study (adapted EQ-5D-Y-3L) to valuation of health states in young children more broadly.

Key findings

1. Use of latent scale DCE to value states in a hypothetical 2-4 year old appears feasible for valuation of the adapted EQ-5D-Y-3L
2. Eliciting preferences from parents/caregivers or those with experience with young children may be beneficial for future valuation studies to generate age-specific value sets
3. Regardless of framing, parents will most often think of their own child while considering their preferences for child health states, although they mentioned that if they had considered a hypothetical child their answers would have been unchanged.
4. Considering and managing the different types of spill-over effects will be crucial for future age-specific valuation studies given that all participants included considerations beyond the task in their answers

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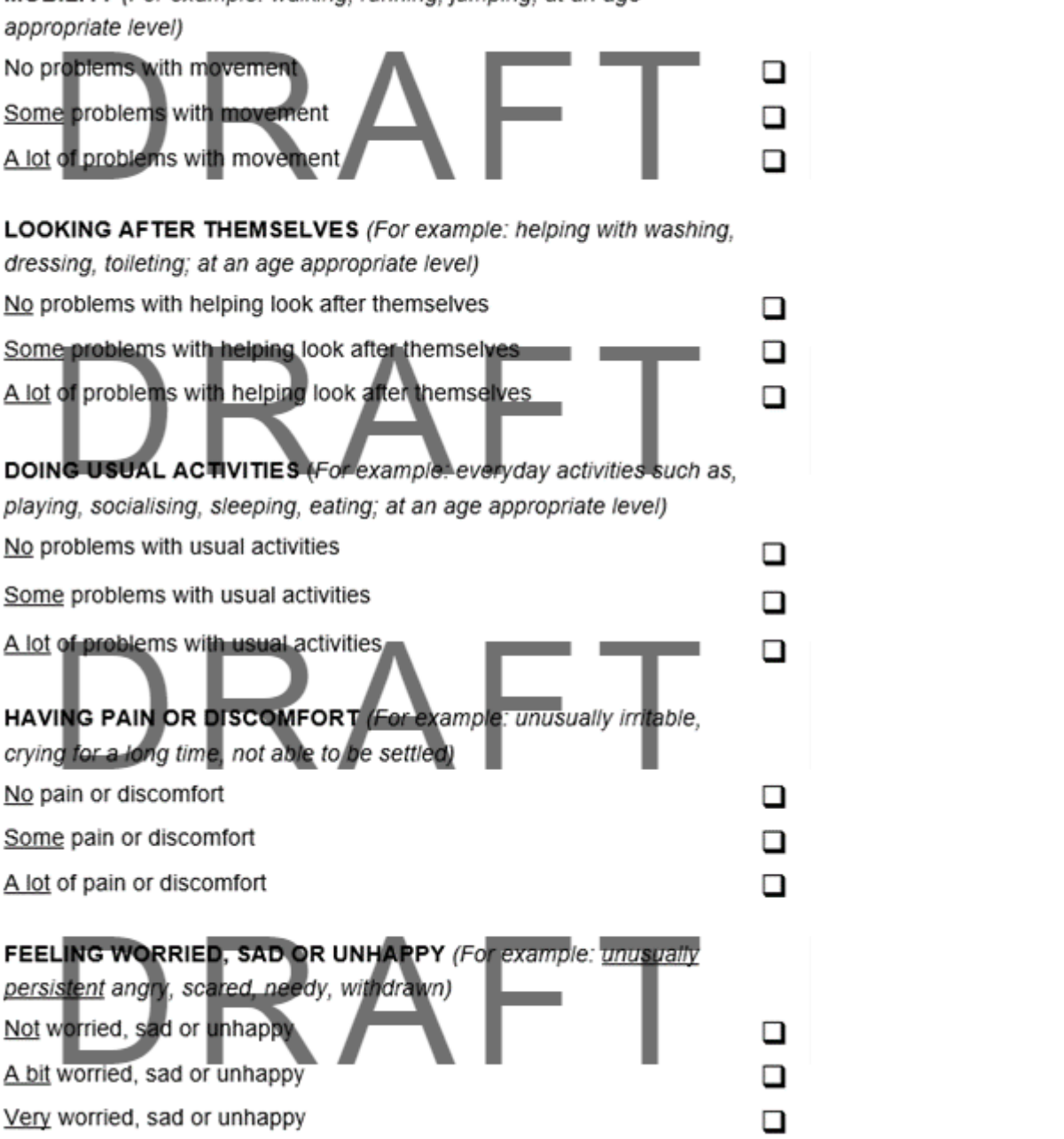
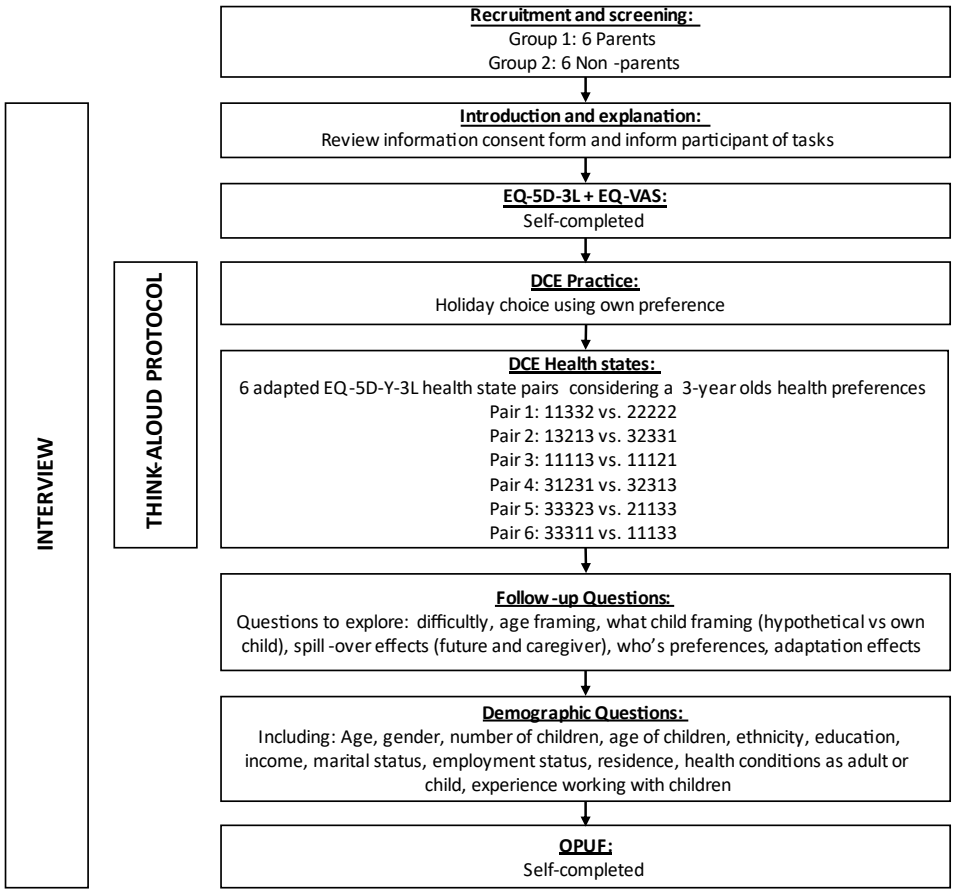
Supplementary material

Supplementary Figure 1 Adapted EQ-5D-Y-3L questionnaire for 2-4 year olds



Supplementary Figure 2 Flow diagram of the study interview



Supplementary Figure 3 Example of a latent scale DCE question

We would like you to complete the exercise while imaging a hypothetical 3-year-old child (not your own child or a child you know). Think about the child's health state TODAY.

Descriptions of health at an age appropriate level	Health State A: Of a hypothetical 3-year-old	Health State B: Of a hypothetical 3-year-old
Mobility: For example: *Walking, running, jumping; at an age appropriate level*	**No** problems with movement	**Some** problems with movement
Looking after self: For example: *Helping with washing, dressing, toileting; at an age appropriate level*	**No** problems with helping look after themselves	**Some** problems with helping look after themselves
Usual Activities: For example: *Everyday activities such as, playing, socialising, sleeping, eating; at an age appropriate level*	**A lot** of problems with usual activities	**Some** problems with usual activities
Pain or discomfort: For example: *Unusually irritable, crying for a long time, not able to be settled*	**A lot** of pain or discomfort	**Some** pain or discomfort
Worried, sad, or unhappy: For example: *Unusually persistent angry, scared, needy, withdrawn*	**A bit** worried, sad or unhappy	**A bit** worried, sad or unhappy

Which do you prefer or which do you think is better? Health State A / Health State B