SYSTEMATIC REVIEW

Preference Elicitation Techniques Used in Valuing Children's Health-Related Quality-of-Life: A Systematic Review

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Abstract

Background and Objectives Valuing children's health states for use in economic evaluations is globally relevant and is of particular relevance in jurisdictions where a cost-utility analysis is the preferred form of analysis for decision making. Despite this, the challenges with valuing child health mean that there are many remaining questions for debate about the approach to elicitation of values. The aim of this paper was to identify and describe the methods used to value children's health states and the specific issues that arise in the use of these methods.

Methods We conducted a systematic search of electronic databases to identify studies published in English since 1990 that used preference elicitation methods to value child and adolescent (under 18 years of age) health states. Eligibility criteria comprised valuation studies concerning both child-specific patient-reported outcome measures and child health states defined in other ways, and methodological studies of valuation approaches that may or may not have yielded a value set algorithm.

Results A total of 77 eligible studies were identified from which data on country setting, aims, condition (general population or clinically specific), sample size, age of respondents, the perspective that participants were asked to adopt, source of values (respondents who completed the preference elicitation tasks) and methods questions asked were extracted. Extracted data were classified and evaluated using narrative synthesis methods. The studies were classified into three groups: (1) studies comparing elicitation methods ($n = 30$); (2) studies comparing perspectives ($n = 23$); and (3) studies where no comparisons were presented ($n = 26$); selected studies could fall into more than one group. Overall, the studies varied considerably both in methods used and in reporting. The preference elicitation tasks included time trade-off, standard gamble, visual analogue scaling, rating/ranking, discrete choice experiments, best-worst scaling and willingness to pay elicited through a contingent valuation. Perspectives included adults' considering the health states from their own perspective, adults taking the perspective of a child (own, other, hypothetical) and a child/adolescent taking their own or the perspective of another child. There was some evidence that children gave lower values for comparable health states than did adults that adopted their own perspective or adult/parents that adopted the perspective of children.

Conclusions Differences in reporting limited the conclusions that can be formed about which methods are most suitable for eliciting preferences for children's health and the influence of differing perspectives and values. Difficulties encountered in drawing conclusions from the data (such as lack of consensus and poor reporting making it difficult for users to choose and interpret available values) suggest that reporting guidelines are required to improve the consistency and quality of reporting of studies that value children's health using preference-based techniques.

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Key Points

We investigated methods used to value children's health states and the specific considerations required in the use of these methods through a systematic review of the literature.

Studies included in the review used a range of preference elicitation methods such as standard gamble, time-trade off, discrete choice modelling, best-worst scaling and visual analogue scales, with and without modification, for different sources of values (who was asked, children or adults or both) and perspectives (point of view that participants were asked to consider).

Deficiencies in reporting made it difficult to compare studies; we recommend the development of guidelines for the design, conduct and reporting of studies in this area.

1 Introduction

There is worldwide interest in how resource allocation decisions can be made around interventions that affect children, and the need to assess the value of the health outcomes they provide in a way that allows for a robust economic evaluation using a cost-utility analysis [1]. Patient-reported outcome measures (PROMs) are widely accepted and used in adult populations for directly measuring health status, quality of life and health-related quality of life (HRQoL) [2]. There are specific challenges, though, in valuing HRQoL for children and adolescents, an important requirement for an economic evaluation using a cost-utility analysis [1, 3]. There are ethical challenges in asking children to engage in valuation tasks, particularly tasks that allow for anchoring of values on a full health to dead scale, as is required for estimation of quality-adjusted life-years (QALYs). Preference elicitation methods need to accommodate the different ages and stages of children's development, and take account of differences in the ways that children perceive their health and health outcomes compared with adults [1].

Elicitation techniques are defined here as the use of stated preference tasks to obtain weights (often referred to as utilities, values or QALY weights) for health states, where health states might be defined by bespoke health state descriptions or vignettes or by the items (dimensions and levels) measured in HRQoL instruments [2]. The term 'values' will be used throughout to encapsulate preference weights (often referred to as utilities, values or QALY weights). The ways

in which values can be derived varies and may include time trade-off (TTO), standard gamble (SG), visual analogue scale (VAS), rating/ranking, discrete choice experiments (DCEs), best-worst scaling (BWS) and willingness to pay (WTP) elicited through contingent valuation methods (referred simply as WTP from here) [2]. Time trade-off and SG can be modified to take quite different forms, and DCEs can be designed to provide only latent scale values or, with the addition of duration as an attribute or by including comparisons with 'dead', to allow the weights to be anchored on a full health to dead scale (where full health has a value of 1 and dead has a value of 0). The techniques of TTO and SG ask participants to value health states by observing their willingness to trade quality of life with duration or risk, respectively; whereas, DCE and BWS techniques ask participants to choose between two or more multi-attribute scenarios, each describing a health state and to make a judgement between them based on preferences. When valuing adult health states, adults are typically asked to value based on their own perspective (i.e. how they would perceive the trade-offs for themselves in that health state, even if it is a hypothetical health state). The additional complexity with valuing children's health is that the respondent not only has to imagine the health state, but may also need to imagine experiencing that health state at a different age and then undertake the valuation [4]. Thus, there are both practical and normative judgements involved in asking adults or children/adolescents to value child/adolescent health. Perspectives may include adults imagining themselves as a child experiencing the health state, imagining their own child (as a parent) or imagining a hypothetical child. Additionally, the age of the imagined child may vary if not specified (which can affect perceptions and preferences). We do not know how valuation differs across the spectrum of a hypothetical child's age or how values change with variation in childhood age.

A range of PROMs has been developed for use in children and adolescents, and value sets have been produced for some of these from which utility values can be derived [5]. Patient-reported outcome measures with value sets for children include the 16D [6], the 17D [7], the Adolescent Health Utility Measure (AHUM) [8], the adolescent version of the Assessment of Quality of Life (AQoL-6D Adolescent) [9], the Child Health Utility instrument (CHU9D) [10], the youth version of the EQ-5D (EQ-5D-Y) [11], the Health Utilities Index Mark 2 (HUI2) [12] and Mark 3 (HUI3) [13], the Quality of Well Being Self-Administered (QWB-SA) scale [14] and the Infant health-related Quality of life Instrument (IQI) [15], each of which covers different age groups and domains. For health technology assessment guidelines internationally, there are a wide range of recommendations about which PROMs to use in economic evaluations of paediatric interventions [16, 17]. In part, the lack of consensus

about which PROMs are preferred is due to the lack of good comparative psychometric evidence on the performance of the available instruments across disease areas and paediatric populations, and this is an area of ongoing research [18].

Previous reviews have described the measurement characteristics of children's generic multidimensional PROMs accompanied by preference-based value sets [5, 19, 20]; however, information on the elicitation methods and perspectives used to obtain these values is lacking. In addition to these value sets for childhood PROMs, other valuation research includes methodological studies, and studies aimed at valuing specific vignettes or scenarios concerning child health. To date, there has been no review of the valuation methods used across these various study types. The aim of the current review, therefore, was to: (i) identify what elicitation techniques have been used to value children's health states (including the valuation task, the sample, the perspective taken for the task and, where relevant, the age of the child whose health is being valued) and (ii) describe the methodological issues specific to valuing child health states (e.g. due to perspective) that have been explored empirically.

2 Methods

A systematic search strategy was used to identify suitable studies for the review. As sourcing methodological information was the main aim, a "systematic search and review" approach was used [21]. This approach is suitable for understanding what is known on a subject and making recommendations for practice and is combined with a comprehensive search strategy. A detailed protocol was developed and agreed on by the authors prior to the searches and adhered to throughout. PROSPERO registration was published on 19/03/2021 (Registration number: CRD42021236494). We define children and adolescents as persons aged under 18 years. For distinctions between the two terms, we relied on the definitions as stated individually by the included studies. The definition of under 18 years was used in this review as this is a commonly accepted age range in many countries worldwide for a child or adolescent; we acknowledge that this is not the case in some countries.

2.1 Information Sources

Five databases were searched: EMBASE, MEDLINE and PsychInfo via Ovid, Econlit and CINAHL via EBSCO. These databases have previously been recommended as suitable for systematic reviews of economic studies in health [22]. The search strategy was based on the following elements: (1) HRQoL studies and childhood PROMs (*what* was being valued); (2) elicitation techniques (*how* it was valued); and (3) a sample from which values were sought

for childhood and adolescent health states (under 18 years of age) [*who* valued it]. The searches were performed on 15 March, 2021. Search terms are presented in Table S1 of the Electronic Supplementary Material (ESM). Additional records were identified by authors in the course of their everyday work.

2.2 Eligibility Criteria

Papers were included if they were published in English from 1 January, 1990 in a peer-reviewed journal, were experimental studies (quantitative or mixed methods) using preference elicitation methods for child health states (under 18 years of age), and concerned with the valuation of PROMs or addressed methodological questions about preference elicitation and valuation. Studies that included the perspective of a child, the perspective of an adult about a child, any preference-based valuation of a child-specific health-related quality-of-life instrument or vignettes for specific diseases were included. Exclusion criteria were studies not published in English, all studies published prior to 1990, publications not in peer-reviewed journals (such as theses/dissertations, conference presentations, abstract only and grey literature), valuation of adult health states and papers on the measurement but not valuation of PROMs.

2.3 Study Selection and Data Extraction

Studies were selected following the Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) reporting guidelines [23]. Publications were screened using the referencing program Rayyan [24]. Titles and abstracts were screened against the inclusion/exclusion criteria by CB, MH, RR and AS. Papers that potentially met the inclusion criteria were accessed in full text and discussed by at least two of the four reviewers against the inclusion/exclusion criteria. Differences in assessment were resolved through discussion and consensus by the four members of the screening team. Papers excluded at the full-text stage are presented in Table S2 of the ESM. Data extraction was performed for the following categories: study, country, title, aims, condition, sample size and age, whose perspective (the point of view that the participant was asked to consider), whose values (who completed the task), values obtained and methods questions asked. Ten percent of the papers were double extracted. When reported, the utility values (0 dead, 1 full health) were extracted and summarised. In some studies, results were expressed as QALYs without underlying utility values. In these cases, the QALY results were extracted as the basis for comparisons. Extraction of results focussed on data relevant to our stated aims, particularly highlighting methodological issues. Results in relation to a specific condition, instrument or comparison of instruments

have not been reported. A formal quality assessment was not conducted, as the primary purpose was to investigate the features of the approaches and methods based on the methodological details reported in the studies. Whilst some outcome data were extracted and reported, given the heterogeneity of conditions and health states addressed these data do not provide a basis to support one method over another. Furthermore, there are no appropriate frameworks or guidelines that enable a meaningful assessment of the varied study types, designs and outcomes.

2.4 Analysis

Data were analysed through a narrative synthesis. Data were presented in separate tables for three groups of studies: (1) those comparing elicitation methods; (2) those comparing perspectives and (3) those with no comparisons presented. Groups 1 and 2 were not mutually exclusive. These categories were chosen so that we could compare methods (Group 1) and perspectives (Group 2). Within the studies included in Group 2, we compared perspectives in three categories: (a) child/adolescents' own perspective, compared to adult/parent values taking the perspective of the child/adolescent, (b) child/adolescents' own perspective, compared to adult/parents' own perspective and (c) adult/parents' own perspective, compared to adult/parent values taking the perspective of a child/adolescent. Tables were constructed for each group from the extracted data and are presented in Tables S3–S5 of the ESM.

3 Results

Including articles sourced through the databases and additional records identified by the authorship group, and after duplicates were removed, we double screened the titles, abstracts and keywords of 1311 papers (CB, MH, RR). Of these, 110 were retained for full-text screening and 77 were included for data extraction (CB, MH, RR, AS), as shown in Fig. 1. There were 30 studies included in Group 1 (comparisons between different elicitation methods), 23 studies in Group 2 (comparison between perspectives) and 26 studies in Group 3 (papers where a single method, measure or perspective was studied, and no comparisons were made). Two papers met the criteria for both groups 1 and 2 [25, 26]. The PROMs examined included variants of the HUI [12, 27–43], as well as the CHU9D [10, 44–50], the EQ-5D-Y [11, 51–61] and the AQoL-6D [9]. In one paper [46], young adults valued their own health; we included this study because its main aim was to obtain values for re-scaling (anchoring) the CHU9D value set for adolescents.

Fig. 1 Summary of the systematic search using the Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) diagram
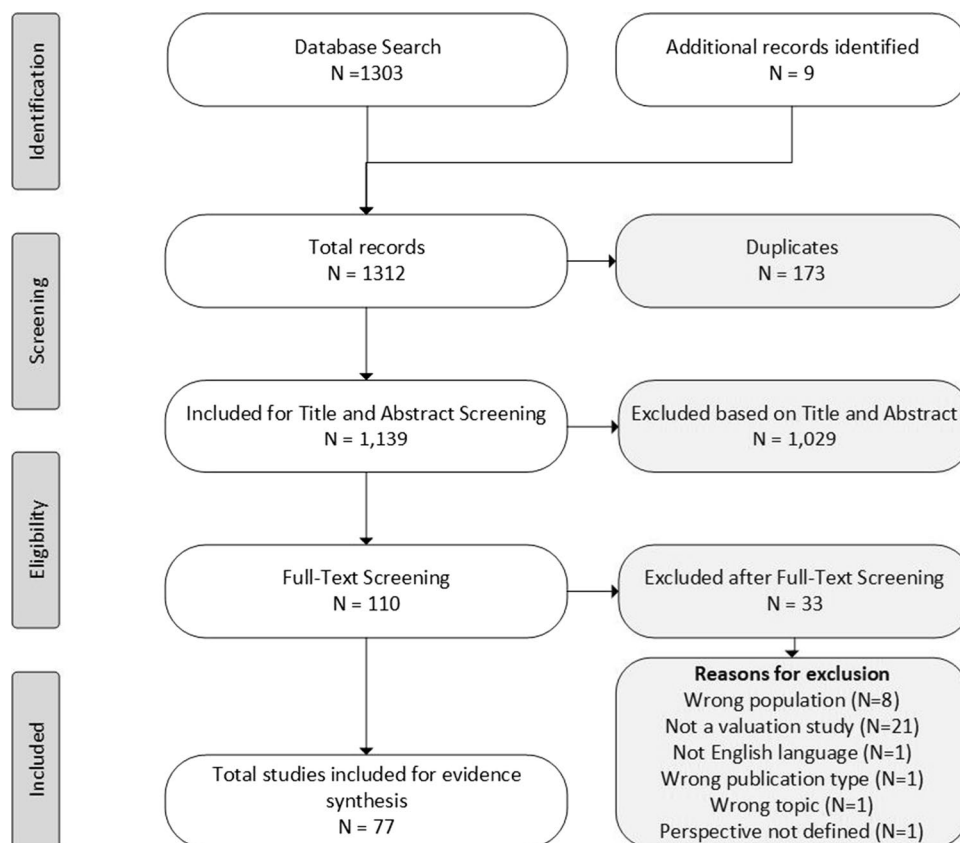
In terms of elicitation methods used across all 77 studies, the application of methods has changed over time as new methods have emerged. Standard gamble has been used regularly since 1996, but thefrequency of use has decreased since a peak in the mid-2000s, while TTO has been used regularly since 2003. Discrete choice experiments first appeared in published reports in this field in 2011 [49] and have become increasingly popular in recent years. Best-worst scaling was first reported in the same paper in 2011 [49], but there have been relatively few papers using BWS since then. Frequency of reporting of the elicitation methods over time is presented in Fig. 2.

3.1 Comparisons Between Different Elicitation Methods (Group 1)

There were 30 studies that compared alternative methods for eliciting preferences for child HRQoL. Table S3 of the ESM contains information on country, study title, aims, health conditions, sample size and age, perspective adopted (the point of view that the participant was asked to consider), source of values (who completed the task) and information on values. Three studies were from Australia [37, 38, 49], seven from Canada [27, 32, 34, 36, 42, 43, 62], one from

Canada and Kenya [59], one from Canada and the USA [41], one from Iran [63], one from Singapore [64], three from the UK [10, 65, 66] and 13 from the USA [26, 28, 39, 67–76]. Elicitation methods used included TTO [26–28, 36, 38, 39, 46, 49, 52, 59, 62–64, 67–72, 74], SG [10, 27, 32, 36, 39, 41–43, 46, 49, 62–64, 67, 69, 72–74, 76], VAS, [26, 27, 32, 36, 38, 39, 41, 42, 52, 59, 62, 63, 67, 69, 73, 76] rating/ranking scale [59], DCEs [49, 52, 65, 68], BWS [49, 65] and WTP [68, 70, 71, 75]. Six (20%) studies [10, 46, 49, 52, 72, 74] were focused on elicitation of values for child health states defined by generic PROMs, with the remainder focused on a range of acute and chronic conditions. The perspective of the studies was highly variable, from consideration of very broad age groups to narrowly defined ages, such as 'a 10-year-old'. Where available, values were extracted for each elicitation method and are presented in Table 1.

In terms of the reporting of the elicitation techniques over time across 30 studies from Group 1, TTO has been used regularly from 1996 to 2021, as have the SG and VAS. Best-worst scaling was used in studies in 2011 [49] and 2021 [65] and DCEs were reported in 2013 [68], 2020 [52] and 2021 [65]. Other than changes in the use of elicitation methods over time, there were no consistent patterns in the reported values between the different elicitation methods. Of note,
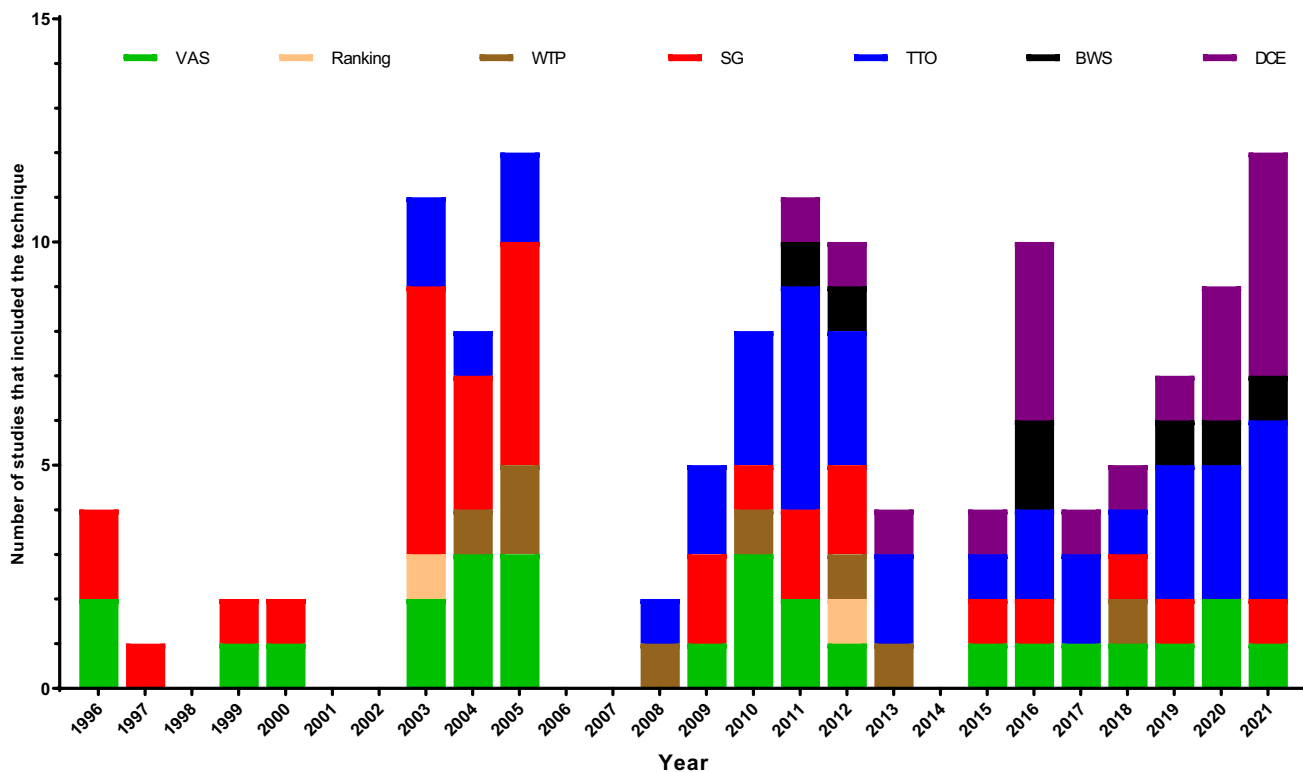

Fig. 2 Frequency count of elicitation methods reported in studies of values for child health-related quality of life between 1996 and 2021. *BWS* best-worst scaling, *DCE* discrete choice experiment, *SG* stand-

ard gamble, *TTO* time trade-off, *VAS* visual analogue scale, *WTP* willingness to pay

there were also variations in approaches taken within each of the elicitation methods, which potentially limits our ability to compare results between methods. One example of this was in three studies in which a modified SG task was presented [27, 36, 73]. In the task, respondents were asked to consider a jar of black and white pills, which represented an instant cure (white pills), and instant painless death (black pills); parents were asked whether they would take a pill from a jar containing a mix of pills with varying proportions of black and white, i.e. changing the probability that selecting a pill will result in instant cure or instant death and give it to their child [36]. Another interesting example was the 'parental TTO', where the poor health state was experienced by the child, but the length of life in full health being traded off was that of the parent [77]. Methodological questions explored in studies included comparisons between elicitation methods [39, 41, 46], whether the order of the valuation technique matters [72], the extent of participant understanding of the technique [65] and comparing conventional SG to a modified SG [36]. There was substantial variation in the application of specific methods, in particular for SG and TTO, including modifications such as avoiding the use of death as an endpoint. In some cases, this reflected the non-fatal nature of the condition being assessed (e.g. tooth decay [65]), in others, it was to limit potential distress for the children participating. [36, 45]

3.2 Comparison Between Perspectives (Group 2)

Twenty-three studies compared values from different perspectives: one from Australia [47], one from Australia and Spain [78], four from Canada [27, 35, 40, 79], two from Europe [54, 55], one from the UK and Europe [80], three from the UK alone [52, 57, 58] and 11 from the USA [25, 26, 28, 31, 67, 77, 81–85]. Table S4 of the ESM contains detailed information on the country, title, aims, target condition, sample size and age, perspective and values for each study.

Thirteen studies included values from the adults' own perspective [28, 47, 52, 54–57, 78, 80–82, 84, 85], nine studies included values from the adolescent's own perspective [26, 31, 35, 47, 78, 81, 82, 84], six studies included values from the child's own perspective [27, 28, 54, 67, 80, 85], and two studies included values from the parent's own perspective [35, 77]. Eight studies included values from the perspective of an adult valuing for the child [25, 52, 55, 56, 58, 78, 83, 85, 86], and four studies included values from the parent valuing for the child, [28, 40, 57, 67]. No studies included values from the perspective of an adult valuing for an adolescent, seven studies included values from the parent valuing for the adolescent [26, 27, 31, 57, 79, 83, 84] and two studies included values from healthcare providers on behalf of children [35, 79]. Health conditions that were

considered for valuation were highly varied, with some samples including participants with specific conditions and some covering health states described by a generic instrument; these differences may have influenced observed differences in values. The elicitation tasks used in the Group 2 studies were predominantly VAS, SG and TTO with only five studies (21%) [47, 52, 58, 78, 81] using a DCE or BWS.

Five studies compared child/adolescent own values with adult/parent values for children (Table 2a). In four of these studies, children/adolescents provided lower values than those provided by adults/parents valuing the child/adolescent health state (type 1 diabetes [28], type 2 diabetes or the risk thereof [31], pelvic inflammatory disease in adolescent girls [26] and generic health states [54]). In only one study in this group (children with cancer) were the child's own values higher than those provided by adult/parent proxies [27].

Of the five studies comparing child/adolescent's own to adult/parents' own values, three studies displayed higher values for adult/parents' own values compared with child/adolescent values (history of extremely low birth weight [35], allergic rhino-conjunctivitis [80] and vision loss [82]). In one study, children estimated their own values higher than adults rated their own values for type 1 diabetes [28], and in one study there was no clear difference between the values of adolescents and adults' own values over a range of health states [47] (Table 2b).

When adult/parents' own values were compared to adult/parent values for the child/adolescent, in three of six studies, adult/parents' own values were higher than adult/parent values for the child/adolescent (type 1 diabetes [57], autism spectrum disorders [77] and congenital differences of sex development [25]). Another three studies found the opposite, with adults' own values being lower than adult values for the child [52, 54, 55] (all three studies investigated a range of health states and were not condition specific). Differences noted in two studies [52, 55] may have been related to the anchoring of the lowest health state (in one of these studies, the differences were only small, and significance was unclear [55]). These two studies also allowed for health states to be considered to be worse than being dead (values less than zero), which may have impacted on the comparability of the values (Table 2c). We were unable to ascertain whether values less than zero were allowed in many of the studies, as shown in Table 2. Eight studies [40, 58, 67, 78, 79, 81, 83, 84] did not fall into any of the above comparison categories (Table 2d).

3.3 Single Elicitation Methods Used, No Comparisons Made (Group 3)

There were 27 papers where no comparisons between elicitation methods or perspectives were made. Of these, three were from Australia [45, 48, 87], three from Canada [12, 33,

Table 1 Study, condition, perspective adopted (who is being asked about), source of values (who is completing task) and values (mean and/or median where information is presented in papers), by year, for Group 1

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and reported values (as presented in papers)
Saigal, 1996 [32]	ELBW	Adolescents (aged 12–16 years) with condition ($n = 149$) and control ($n = 124$)	Adolescents (12–16 years) with condition and control	Adolescents (aged 12–16 years) with condition and control	SG, condition mean (SD) = 0.87 (0.26); median = 1.00; range = −0.5 to 1.00 Control mean (SD) = 0.93 (0.11); median = 0.95; range = 0.55–1.00 VAS, condition mean (SD) = 87 (20); median = 94; range = −1 to 100 Control mean (SD) = 91 (8); median = 93; range = 46–100
Brunner, 2003 [41]	Chronic MSKDs	Parents of children with MSKDs and children aged 8 years or older ($n = 55$)	Child, parents were asked to complete the questionnaires as they thought their children would	Parents of children aged 1–18 years with MSKDs and children aged over 8 years with MSKDs	SG, parents: mean = 0.50–1.0; median = 0.935–0.995 Child: mean = 0.45–1.0, median = 0.881–0.915 Linear analogue scale, parents: mean = 0.09–1.0, median = 0.777–0.775 Child: linear analogue scale: mean = 0.18–0.99, median = 0.691–0.695 Categorical scale, parents: mean = 0.17–1.0, median = 0.710–0.667 Child: categorical scale: mean = 0.17–1.0, median = 0.630–0.667 HUI3, parents mean = − 0.10–1.0, median = 0.762–0.864 Child: HUI3: mean = 0.17–1.0, median = 0.796–0.863
Saigal, 2003 [34]	ELBW	Adults Antenatal cohort of women with high-risk pregnancies ($n = 81$) Mothers of very low birth weight children ($n = 90$)	Self, imagine living in the health state for the next 60 years	Mothers	Focus of study was on stability of values. VAS and SG used, but unclear how values were derived
Saw, 2003 [64]	Myopia	Students with myopia aged 15–18 years ($n = 699$)	Self	Teenagers	TTO, mean (95% CI) = 0.93 (0.93–0.94); median = 0.97 SG, mean (95% CI) = 0.85 (0.84–0.86); median = 0.95

Table 1 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and reported values (as presented in papers)
Sung, 2003 [36]	Cancer	Parents mean age 39.5 SD = 6.3 (n = 85)	Self, own child for adults	Parents and children	TTO, oncology patients: mean (SD) = 0.64 (0.33) Non-oncology out-patients: mean (SD) = 0.88 (0.14) SG, oncology patients: mean (SD) = 0.83 (0.29) Non-oncology out-patients: mean (SD) = 0.93 (0.12) Modified SG, oncology patients: mean (SD) = 0.92 (0.23) Non-oncology out-patients: mean (SD) = 0.96 (0.11) VAS, oncology patients: mean (SD) = 0.71 (0.22) Non-oncology out-patients: mean (SD) = 0.81 (0.18) HUI2, oncology patients: mean (SD) = 0.85 (0.18) Non-oncology out-patients: mean (SD) = 0.80 (0.19) HUI3, oncology patients: mean (SD) = 0.84 (0.23) Non-oncology out-patients: mean (SD) = 0.76 (0.24)
Yi, 2003 [39]	Cystic fibrosis	Adolescents with cystic fibrosis aged from 12 to 18 years (n = 65)	Self	Adolescents	TTO, mean (SD) = 0.96 (0.07) SG, mean (SD) = 0.92 (0.15) VAS, mean (SD) = 0.76 (0.20) HUI2, mean (SD) = 0.83 (0.16)
Brunner, 2004 [73]	Chronic arthritis in children	Families of children (n = 119; 91 girls and 28 boys) with chronic arthritis. The mean age of the enrolled children was 10.5 years (range 3–18 years; SD 4.3 years)	Child aged 3–18 years	Parents of children aged 3–18 years (12 fathers, 106 mothers and 1 grandmother) and children aged 8 years or more	Modified SG, child: mean = 0.89, median = 0.94 Modified SG, all parents: mean = 0.91, median = 0.96 Modified SG, parents with child: mean= 0.89, median =0.94 VAS, child: VAS-health: mean = 7.7, median =8.1 VAS, all parents: VAS-health: mean = 8.0, median = 7.5 VAS, parents with child: VAS-health: mean = 7.3, median = 7.6

Table 1 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and reported values (as presented in papers)
Feeny, 2004 [43]	ELBW	Own perspective of health states, imagine it for the rest of their life. VAS and HUI for their own usual health state	Child	Adult parents (aged > 18 years) of children as proxy for their child	SG, ELBW mean (SD) = 0.90 (0.20), median = 1.00 SG, control mean (SD) = 0.93 (0.11), median = 0.95 HUI2, ELBW mean (SD) = 0.89 (0.14), median = 0.95 HUI2, control mean (SD) = 0.95 (0.09), median = 1.00 HUI3, ELBW mean (SD) = 0.80 (0.22), median = 0.87 HUI3, control mean (SD) = 0.89 (0.13), median = 0.93
Prosser, 2004 [70]	Illnesses/outcomes preventable by pneumococcal conjugate vaccine	Adults: parents of children who have experienced outcomes (n = 101) and general population (n = 109)	Newborn	Adults	TTO, median days given up to avoid health states (not transformed to values): parents = 0–270; community = 0–365 WTP, median WTP: parents = US$25–500; community = US$50–500
Sung, 2004 [27]	Cancer	Parents mean 43 SD 5 (n = 22) Children mean 14 SD 2 (n = 22)	Self, own child and hypothetical scenarios	Parents and children	Parent, mean SG = 0.92 VAS = 0.76 TTO = 0.77 HUI2 = 0.82 HUI3 = 0.79 Child, mean (SD) SG = 0.92 VAS = 0.80 TTO = 0.92 HUI2 = 0.95 HUI3 = 0.92
Chiou, 2005 [76]	Asthma	Adults (n = 124)	Child	Adults, general population	SG, mean = 0.06–0.96 VAS, mean = 0.03–0.79
Prosser, 2005 [75]	Influenza	Adults (n = 112)	Child	Adult	TTO, median 0 days to 3 years, mean 68 days to 5 years (not transformed into values) WTP, median: US$100–5000, mean: US$288–28,579

Table 1 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and reported values (as presented in papers)
Chen, 2008 [71]	Acne	Children aged 14–18 years (n = 216)	Self for TTO by adolescents only, Adolescent for WTP for parent	Adolescents for TTO, parents for WTP	TTO, adolescents 100% clear: mean = 0.978; median = 0.994 50% clear: mean = 0.967; median = 0.992 100% clear with scarring: mean = 0.965; median = 0.992 Current state: mean = 0.961; median = 0.985 WTP, adolescents: Never had: median = $275 100% clear: median = $100 50% clear: median = $10 100% clear with scarring: median = $0 WTP, parents of adolescents: Never had: median = $275 100% clear: median = $100 50% clear: median = $100 100% clear with scarring: median = $0
Carroll, 2009 [74]	Multiple health states representative of a range of conditions	Men and women aged over 18 years, with at least 1 child aged under 18 years at the time of the study (n = 4016)	Child (assume aged 1–18 years)	Parent	TTO, mean = 0.51–1.00, median = 0.50–1.00, mode = 0.50–1.00, SG, mean = 0.59–1.00, median = 0.50–1.00, mode = 0.50–1.00 (range presented to cover multiple outcome values)
Yi, 2009 [69]	IBD	Adolescents with (n = 67) and without (n = 88) IBD	Self	Adolescents; own preferences	TTO, with IBD mean (SD) = 92.3 (17.2); without IBD mean (SD) = 98.5 (2.9) SG, with IBD mean (SD) = 97.1 (7.5); without IBD mean (SD) = 98.4 (3.4) VAS, with IBD mean (SD) = 77.6 (16.2); without IBD mean (SD) = 86.7 (8.4)
Lee, 2011 [28]	Type 1 diabetes mellitus	Adults (aged >18 years) [n = 213], children (aged 8–18 years) [n = 238], and adult (aged >18 years) parent proxy for children (aged 8–18 years) [n = 223]	Adults: self Children: self Adult parent: their child	Adults, children, adult parent proxy	HUI, mean (SD) = 0.85 eTTO, mean (SD) = 0.81

Table 1 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and reported values (as presented in papers)
Ratcliffe, 2011 [49]	Generic health states	School children (n = 16)	Self	Adolescents aged 11–13 years	CHU9D, mean = 0.85 Scores for TTO and SG not presented
Trent, 2011 [26]	Pelvic inflammatory disease	Adolescent girls (n = 134) mean 16 (1.7) and parents of adolescents (43 (10)	15-year-old adolescent girl Adults, their child	Adolescents, self Adults and parents of adolescents	TTO, range of PID health states: mean (SD) = 0.85–0.902 (0.25–0.31); median 0.98–1.00 VAS, range of PID health states: mean (SD) = 60.87–76.24 (23.33–27.05); median 60.0–85.0 (range presented to cover multiple outcome values)
Finnell, 2012 [72]	Set of 29 generic paediatric health states	Adult parents (aged >18 years) of children as proxy for their child	Child	Adult parents (aged >18 years) of children as proxy for their child	TTO, 29 health states: mean 0.45–0.97 SG, 29 health states: mean 0.58–0.97
Stevens, 2012 [10]	Generic health states	Adults (n = 282), general population aged mean 49 years	Self, asked to consider being the health state for the rest of their lives	Adults	Mean (SD) over range of health states = 0.9316–0.3369 (0.1027–0.3154); median 0.975–0.45 (paper used SG and ranking, but it is unclear how values were calculated)
Tong, 2013 [38]	Chronic kidney disease	Adolescents and young adults with chronic kidney disease age range 12–25 years (mean 17 years) [n = 27]	Self	Adolescents and young adults	TTO, mean (SD) = 0.99 (0.001) VAS, mean (SD) = 0.83 (13.8) [assume that the SD was not converted to 0–1 scale] HUI2, mean (SD) = 0.863 (0.164) HUI, mean (SD) = 0.854 (0.212)

Table 1 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and reported values (as presented in papers)
Prosser, 2013 [68]	Influenza	Adults (n = 1012)	Hypothetical individual aged 1, 3, 8, 15, 35, 55, 70 and 85 years	Adult (aged >18 years)	Marginal TTO, mean (95% CI) days traded uncomplicated influenza = 16.34 (9.63–25.06); hospitalisation day averted = 9.63 (6.68–13.33) Marginal time traded DCE, mean (95%CI) days traded uncomplicated influenza = 14.89 (9.97–20.33); hospitalisation day averted = 26.62 (13.98–40.94) WTP, mean (95% CI) days traded uncomplicated influenza = 66.27 (30.02–131.64); hospitalisation day averted = 168.26 (126.24–206.88) WTP DCE, mean (95% CI) days traded uncomplicated influenza = 62.54 (46.98–77.89); hospitalisation day averted = 513.06 (223.52–878.82) (not transformed into utility values)
Ratcliffe, 2015 [46]	Generic health states	18- to 29-year-old university students (n = 152)	Self	Young adults	TTO, mean = 0.63 to − 0.021; median = 0.65 to 0 SG, mean = 0.83 to 0.33 (range presented to cover multiple outcome values)
Ebrahim, 2015 [42]	Paediatric intensive care unit	Parents of 51 children aged ≥ 4 years	Own child	Adults	SG, converted SG score: N = 51; baseline mean (SD) = 0.82 (0.19); 1-month mean (SD) = 0.81 (0.15) VAS, VAS rating: N = 51; baseline mean (SD) = 72.53 (25.36) mm; 1-month mean (SD) = 69.97 (22.03) mm HUI3: N=52; baseline mean (SD) = 0.70 (0.39); 1-month mean (SD) = 0.58 (0.39)

Table 1 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and reported values (as presented in papers)
Dillman, 2016 [67]	Newly diagnosed small bowel Crohn disease	Children (age range = 9–18 years) [n = 26] Parent/guardian of the 26 children as proxy	Self for children; parent/guardian, their child	Children and adults	TTO, before: median (IQR) = 7.7% (6.2–15.4%), after: (IQR) = 7.7% (6.2–15.4%), $N = 21$, $p = 0.32$ SG, before: median (IQR) = 90.0% (80.0–95.0%), after: (IQR) = 91.0% (75.0–95.0%), $N = 21$ $p = 0.65$ VAS, before: median (IQR) = 47.5 (20.0–52.2), after: (IQR) = 83.0 (62.0–92.0, $N = 21$, $p = 0.0003$
Poenaru, 2017 [59]	15 health states of key paediatric congenital anomalies	Adults, health professionals and community care givers ($n = 154$)	Child	Adult health professional or caregiver	Ranking, Kenya: 0.20–1.00; Canada: 0.067–0.933 VAS, Kenya: 0.20–0.81; Canada: 0.185–0.885 TTO, Kenya: 0.126–0.748; Canada: 0.037–0.773 PC-TTO, Kenya: 0.212 to 0.758; Canada: 0.055–0.797 PC-Global Burden of Disease, Kenya: 0–0.834; Canada: 0–0.812
Shahjouei, 2019 [63]	Tethered spinal cord and prone positioning	Mothers aged 30.5 years, SD = 6.7 ($n = 74$)	Own child	Mothers	TTO, mean (SD): TTO = 0.68 (0.36); m-TTO = 0.66 (0.39) SG, mean (SD) 0.38 (0.43); chain of gambles 0.68 (0.36) VAS2a, mean (SD): = 0.13 (0.57) VAS2b = 0.57 (0.28) VAS3a = 0.17 (0.55) VAS3b = 0.58 (0.27)

Table 1 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and reported values (as presented in papers)
Shah, 2020 [52]	Generic health states	General public, (n = 299), broadly reflecting age and sex for UK population	Adult for EQ5D, Child for EQ5D-Y	General population	TTO, used for anchor for state 33333, values using the child perspective higher than adult perspective (data not given) VAS, used for anchor for state 33333, values using the child perspective higher than adult perspective (data not given) DCE EQ-5D-3L adult perspective: 0.392–0.633; child perspective: 0.313–0.653. EQ-5D-Y adult perspective: 0.167–0.667; child perspective: 0.167–0.667
David, 2021 [62]	Ameloblastoma of the lower jaw	Adults in a university setting (mean age 29.9– years) [n = 86]	Adult asked to imagine themselves as the pictured 10-year-old child with a voluminous ameloblastoma of the lower jaw	Adults	TTO, mean (SD) 0.65 (0.22) SG, mean (SD) 0.64 (0.20) VAS, mean (SD) 0.60 (0.17)
Rogers, 2021 [65]	Dental caries (tooth decay)	Children aged 11–16 years (n = 33)	Self	Children aged 11–16 years	DCE, values not presented BWS, values not presented

SD and other variance measures are included in the ESM and Lavelle 2019 appears in two sections

ALL acute lymphoblastic leukemia, *BWS* best-worst scaling, *CHU9D* Child Health Utility 9 Dimensions, *CI* confidence interval, *cTTO* composite time-trade off, *DCE* discrete choice experiment, *ELBW* extremely low birth weight, *HUI2* Health Utility Index Mark 2, *HUI3* Health Utility Index Mark 3, *IBD* inflammatory bowel disease, *LOD* location of dead, *MSKDs* musculoskeletal disorders, *PC* paired comparison, *QALY* quality-adjusted life-year, *SD* standard deviation, *SG* standard gamble, *TTO* time trade-off, *VAS* visual analogue scale, *WTP* willingness to Pay

Table 2 Study, condition, perspective adopted (who is being asked about), source of values (who is completing task?), elicitation methods and measures and values (mean and/or median where information is presented in papers) presented in four sections for comparison of perspectives, by year, for group 2

a) Comparison of values: child/adolescents' own to adult/parent values for child/adolescent (studies are bolded where child/adolescent's own values are lower than parent values for child/adolescent)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers) by perspective	Summary of findings for comparison of values	Values less than zero allowed?
Sung, 2004 [27]	In patients with cancer vs outpatients without cancer	Parents mean age 43 years; Children mean age 14 years (*n* = 27)	Self for children, own child for parents (health states from HUI2/3)	Parents and children	SG, VAS, HUI	Child, self Mean: SG: 0.92; VAS 0.80; TTO: 0.92; HUI2: 0.95; HUI3: 0.92 Parent values for child Mean: SG: 0.92; VAS 0.76; TTO: 0.77; HUI2: 0.82; HUI3: 0.79	Child's own values are higher than parent values for child	Unclear (no values less than zero presented)
Lavelle, 2011 [85]	Pandemic influenza A (H1N1) illness and vaccine-related adverse events	Parents of children aged 3–17 years diagnosed with ASD (ASD group) and parents of children aged 3–17 years with no diagnosis with ASD (comparison group) [*n* = 255]	Family member or friend matching the ages (1, 8, 35 and 70 years) for hypothetical health states	Adults	TTO, WTP, QALY	QALY loss Adult, self Age 35 years, mean = 0.0138 to 0.0300 Age 70 years, mean = 0.0074 to 0.0135 Adult values for child Age 1 year, mean = 00317 to 0.0475 Age 8 years, mean = 0.0281 to 0.0391	Higher QALY loss for adult values for children than adults own values (i.e. lower QALYs for children than proxy) [note reported as QALYs not utilities]	Unclear (trading off days only, no values less than zero presented)
Lee, 2011 [28]	T1D	Adults (aged >18 years) [*n* = 213], children (aged 8–18 years) [*n* = 238], and adult (aged >18 years) parent values for children (aged 8–18 years) [*n* = 223]	Adults, self Children, self Adult parent, own child	Adults, children, adult parent values for children	TTO, HUI3	Child, self HUI3 mean = 0.89 eTTO mean = 0.81 Adult, self HUI3 mean = 0.85 eTTO mean = 0.81 Parent values for child HUI3 mean = 0.91 eTTO mean = 0.84	Child's own values are lower than parent values for child	Unclear (no values less than zero presented)
Rhodes, 2011 [31]	Type 2 diabetes or at risk	Parent (*n* = 66) and adolescent (aged 12–18 years) [*n* = 65]	Adolescents, self Parent own child (adolescent)	Parents and adolescents (aged 12–18 years) with/at risk of type 2 diabetes	SG, HUI3	SG (HUI3 not presented) Adolescent, self Range of median = 0.51–0.82 Parent values for adolescent Range of median = 0.80–1.0	Adolescent's own values are lower than parent values for adolescent	No

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Table 2 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers) by perspective	Summary of findings for comparison of values	Values less than zero allowed?
Trent, 2011 [26]	Pelvic inflammatory disease	Adolescent (aged 12–19 years) girls ($n = 134$) and parents of adolescents ($n = 121$)	All participants asked to consider a 15-year-old girl with PID. Adolescents, self t Parents, their child	Adolescents and parents of adolescents	TTO, VAS	Adolescent, self Mean range 0.76–0.84; median range 0.98–1.00 Parent values for adolescent Mean range 0.85–0.90; median range 0.98–1.00	Adolescent's own values are lower than parent values	Unclear (no values less than zero presented)

b) Comparison of values: child/adolescents' own to adult/parents' own (studies are bolded where child/adolescent's own values are lower than adult/parents' own values)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers) by perspective	Summary of findings for comparison of values	Values less than zero allowed?
Saigal, 1999 [35]	Extremely low birth weight	Healthcare professionals Parents of extremely low birth weight and normal weight ($N = 478$)	Preferences for 4 or 5 generic health states in children considered by healthcare professionals, child; adolescents, self; parents, their child	Healthcare professionals and parents	SG, VAS, health states based on HUI2	Adolescent, self Mean range = 0.16–0.72 Parent, self Mean range = 0.20–0.82 Healthcare professionals Nurses: mean range = − 0.07 to 0.79 Physicians: mean range = − 0.03 to 0.86	Adolescents' own values lower than parent values	Yes
Lee, 2011 (also in previous group) [28]	T1D	Adults (aged >18 years) [$N = 213$], children (aged 8–18 years) [$N = 238$], and adult (aged >18 years) parent values for children (aged 8–18 years) [$N = 223$]	For TTO, generic health states, with varying life expectancy depending on participants' age Adults: self. Children: self. Adult parent: their child	Adults, children, adult parent values for child	TTO, HUI3	Child, self HUI3 mean = 0.89 eTTO mean = 0.81 Adult, self HUI3 mean = 0.85 eTTO mean = 0.81 Parent values for child HUI3 mean = 0.91 eTTO mean = 0.84	Child self-values are higher than adult self on the HUI3, but not the eTTO	Unclear (no values less than zero presented)
Ratcliffe, 2016 [47]	Generic health states	Adults aged >18 years ($n = 625$)	Adults aged >18 years asked to rate CHU9D (child) health states (included as aim to develop new adolescent-specific scoring)	Adults aged >18 years	BWS-DCE, Child Health Utility 9D	Adolescent, self Adolescent BW value range 0.769–0.856 Adult, self Adult BW value range 0.734–0.886	Adolescent's values range is more constrained than adults', no clear direction	Unclear (no values less than zero presented)

Table 2 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers) by perspective	Summary of findings for comparison of values	Values less than zero allowed?
Retzler, 2018 [80]	Allergic rhino-conjunctivitis	Adults and children aged 8–11 years (n = 1454)	Adult and child, self Vignettes of health states related to allergic rhino-conjunctivitis	Adult and child	SG (adult) and VAS (child)	Child, self Values range 0.635–0.705; median range 0.610–0.705. Adult, self Values range 0.812–0.880; median range 0.851–0.967	Child's own values are lower than adults', (methodology different, SG for adult, VAS for child)	Unclear (no values less than zero presented)
Stevens, 2021 [82]	Vision loss	Secondary students and recent graduates, mean 16.8 (SD 1.2) [n = 145]	Youths and adults, self Vignettes about vision loss	Youths aged 13–20 years and adults from the general community from previous study	TTO, maximum amount of remaining life they would trade for an intervention that guarantee perfect bilateral vision	Adolescent, self Youth cohort mean range 0.79–0.96 Adult, self Patients with vision loss: mean range 0.60–0.78 Adult general community mean range 0.86–0.96	Adolescents' own values are lower than adults' in the general community	Unclear (no values less than zero presented and vision loss unlikely to be seen as worse than death)

c) Comparison of values: adult/parents' own to adult/parent values for child/adolescent (studies are bolded where adult/parents' own values are higher than adult/parents' values for child/adolescent)

Table 2 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers) by perspective	Summary of findings for comparison of values	Values less than zero allowed?
Lloyd, 2010 [57]	T1DM	Adults (aged ≥ 18 years) general public (n = 100), adults (aged ≥ 18 years and <35 years) with T1DM (n = 51), parents of children (aged 8–12 years), adolescents (aged 13–17 years) with T1DM (n = 20)	Adult general public, self. Adults with T1DM, self. Adult parents of children (aged 8–12 years): their child. Adult parents of adolescents (aged 13–17 years): their child Vignette descriptions of T1DM health states	Adults, children, adult parent values	VAS, SG, EQ-5D	Adult, self, general public Mean EQ-5D VAS = 81.7 EQ5D single index, mean = 0.921 SG, mean at base range = 0.976 Adults with T1DM EQ-5D VAS, mean = 78.8 EQ-5D single index, mean = 0.886 SG, mean at base range = 0.938 Parent as if they were a child with T1DM EQ-5D VAS, mean = 80.5 EQ5D single index, mean = 0.840 SG, mean at base range = 0.947 Parent as if they were adolescent with T1DM EQ-5D VAS, mean= 80.8 EQ-5D single index, mean = 0.765 SG, mean at base range = 0.955	Adults' own values are higher than parent values for child and adolescent with T1DM on the EQ5D and SG, and slightly higher for the VAS	No, SG and VAS constrained to death as 0 with no negative values presented

Table 2 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers) by perspective	Summary of findings for comparison of values	Values less than zero allowed?
Kind, 2015 [54]	8 EQ-5D-Y generic health states	Adults (n = 1085)	Self, other adult and 10-year-old child (n = 542) Set of EQ-5D-Y health states	Adults	VAS, EQ-5D-Y	Mean values for EQ-5D-Y health states Adult, self Germany 15.417–81.202 Spain 20.452–86.947 England 20.012–79.812 Adult, other Germany 19.798–85.639 Spain 33.491–88.895 England 25.057–87.394 Adult, values for child Germany 21.017–86.055 Spain 23.833–88.276 England 20.972–82.185	Adults' own values are lower than adult values for child	No, VAS scores constrained to 0–100
Kreimeier, 2018 [55]	Generic health states	Adults (n = 805)	Adult self and; 10-year-old child. For set of EQ-5D-Y health states	Adults	C-TTO/DCE + death; EQ-5D-Y; EQ-5D-3L	Adult, self EQ-5D-3L mean range − 0.32 to 0.94 EQ-5D-Y mean range − 0.17 to 0.95 Adult values for child EQ-5D-3L mean range − 0.20 to 0.94 EQ-5D-Y mean range − 0.14 to 0.96	Adult levels slightly lower at lowest range (33333)	Yes, anchor at death appears to be interpreted differently between adults and children
Lavelle, 2019 [77]	Autism spectrum disorders	Adult community members (n = 718)	Parent's, self and their child, and trading own life for both their child and adult health	Parents	TTO	Parent values for child Mean = 0.49–0.80 Parent, self Mean = 0.75–0.93 Composite (both) Mean = 0.45–0.78	Adults' own values higher than parent values for child	Unclear (no values less than zero presented)

Table 2 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers) by perspective	Summary of findings for comparison of values	Values less than zero allowed?
Alkazemi, 2020 [25]	Congenital differences of sex development	Adults, general population (n = 1647)	6-year-old with 10 years left to live (1) Adult asked to trade off a 6-year-old's life with 10 years yet to live (2) Combined adult and 6-year-old's expected years left to live traded off to get the child back to perfect health	Adults, general population	TTO	Adults trading off child's life (values for child) vs trading off child AND adult's life (age 6 years, 10 years to live) [used here as values for adult.] Adult values for child, mean = 0.67; median = 0.70 Combined life years: mean = 0.80; median = 0.88	Adults' own values (using values of combined years) are higher than adults values for the child	Unclear (no values less than zero presented)
Shah, 2020 [52]	Generic health states	General public (n = 299), broadly reflecting age and sex for UK population	Self, adult EQ-5D-3L health states Child for EQ-5D-Y health states	General population	TTO, VAS, DCE, LOD (location-of-dead) EQ-5D-3L, EQ-5D-Y	TTO, used for anchor for state 33333, child perspective higher than adult perspective (data not given) VAS, used for anchor for state 33333, child perspective higher than adult perspective (data not given) DCE results Adult perspective EQ-5D-3L, mean = 0.333–0.633 EQ-5D-Y, mean = 0.167–.667 Child perspective EQ-5D-3L, mean = 0.313–0.653 EQ-5D-Y, mean = 0.167–0.667 [note that the EQ-5D-Y range is the same for both perspectives of the EQ-5D-Y, but the values are for different health states]	Adult's own values were lower than adult values for the child for TTO, VAS and LOD but inconclusive for DCE values	Yes, worse than dead values were allowed for TTO

Table 2 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers) by perspective	Summary of findings for comparison of values	Values less than zero allowed?
d) Comparisons other than the above								
Barr, 1997 [40]	Acute lymphoblastic leukaemia	Children receiving 'maintenance' chemotherapy aged 11 months to 14 years (n = 18)	Children, self Adults, child Range of vignettes related to acute lymphoblastic leukaemia	Patients, family members and health professionals	HUI2 and HUI3	Nurse/physician parents about child Mean: Week 1: nurse = 0.96, physician = 0.90, parents = 0.86 Week 2: nurse = 0.86, physician = 0.83, parents = 0.83 Week 3: nurse = 0.91, physician = 0.89, parents = 0.89	Comparison only between healthcare professionals and parents	Unclear (no values less than zero presented)
Wasserman, 2005 [84]	Haemophilia	Children (aged 7 months to 18 years) with haemophilia and adults mean aged 35 years with haemophilia (n = 128)	Adolescents, self Parent, their child health states specific to haemophilia presented with a short description	Adolescents, adults and parents	VAS and SG	Child and adolescents, with parent values for children aged below 13 years VAS, mean range = 0.237–0.926 SG, mean range = 0.487–0.936 Adult, self VAS, mean range = 0.258–0.897 SG, mean range = 0.422–0.884	Cannot compare as parent values are combined with adolescent self-report	Unclear (no values less than zero presented)
Ethier, 2012 [79]	Oral mucositis prevention in paediatric cancer	Parents of children (aged 1'18 years old) [n = 82] receiving intensive chemotherapy for leukaemia/lymphoma or undergoing stem cell transplantation. Healthcare professionals (n = 60) [physicians, nurses or pharmacists] caring for children with cancer]	Parents, their child with cancer HCPs – child	Parents and HCPs	VAS, TTO and WTP	Parent values for adolescent Reduction in survival time with TTO (weeks), median; mild mucositis = 0.0; severe mucositis = 0.0 Healthcare provider Reduction in survival time with TTO (weeks), median; mild mucositis = 0.0; severe mucositis = 3.0	Values not meaningful in this context as parents were not willing to trade time to prevent mild or severe mucositis	Unclear

Table 2 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers) by perspective	Summary of findings for comparison of values	Values less than zero allowed?
Dillman, 2016 [67]	Newly diagnosed small bowel Crohn's disease	Children (age range = 9–18 years) [n = 26]; parent/guardian values for the 26 children	Child, self Parent/guardian, their child. Vignettes relating to mucositis states	Children and adults	VAS, TTO, and SG	Child, self TTO: before: median = 7.7% After: median = 7.7% SG: before: median = 90.0% After: median = 91.0% VAS: before: median = 90.0% After: median = 91.0% Parent values for child Correlations discussed but values not reported	Parent values not presented, outcome unclear	Unclear
Law, 2017 [81]	Generic health states	Adults aged ≥ 18 years (n = 44) Adolescents aged ≥ 12 years and <18 years (n = 55)	Self hypothetical health states loosely inspired from the EQ-5D	Adults own perspective and adolescent's own perspective	DCE, pairwise comparison, Comparison between scenarios (loosely based on EQ-5D)	Adolescent, self Odds ratios but values not presented Adult, self Odds ratios but values not presented	Values not presented	Yes, preferences for death compared to worst health state measured
Tejwani, 2017 [83]	Vesicoureteral reflux	General public, adults (n = 1627) mean age 34.9 years	Parent of hypothetical child 1. Trades from 'their child' in exchange for time disease free for time disease free 2. Trades from own life to benefit their child 3. Dyadic-combined trades from the parents, using vignettes related to vesicoureteral reflux	Adults from general public	TTO	Values not presented	Values not presented	No, negative values not possible in experimental design

Table 2 (continued)

Author, year	Condition	Sample size and age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers) by perspective	Summary of findings for comparison of values	Values less than zero allowed?
Dalziel, 2020 [78]	Set of 242 generic health states produced by combination of EQ-5D-Y domains	Australia: (n = 2134) adults (aged ≥ 18 years) and 1010 adolescents (aged 11–17 years) Spain: (n = 2007) adults (aged ≥ 18 years) and 1000 adolescents (aged 11–17 years)	Adults, self and 10-year-old child. Adolescents Self health states derived from the EQ-5D-Y	Adults and adolescents	BWS, DCE, EQ-5D-Y (EQ-5D-5L)	Marginal frequencies: Adolescent, self Australian adolescents: Mobility1 best at 0.482; Sad/Worried3 worst at 0.438 Spanish adolescents: Pain1 best at 0.508; Pain3 worst at 0.438 Adult, self Australian adults Pain1 best at 0.472; Pain3 worst at 0.495 Spanish adults Pain1 best at 0.506; Pain3 worst at 0.441 Adult values for child Australian adult values: usual activities1 best at 0.493; Pain3 worst at 0.565 Spanish adult values: Pain1 best at 0.548; Pain3 worst at 0.530	Values not calculated as focus was on preferences. Hard to interpret BWS scores Australian adults place less weight on being very worried, sad or unhappy compared with Australian adolescents	Unclear (no values less than zero presented)
Mott, 2021 [58]	Generic health states	Adults from general population (n = 1000), adolescents aged 11–17 years (n = 1005)	Adults, 10-year-old child Adolescents, self Health states from the EQ-5D-Y-3L	Adults and adolescents	DCE, EQ-5D-Y-3L	Values not presented	Values not presented	Not applicable, data to anchor the DCE not collected

SD and other variance measures are included in the ESM and Lavelle 2019 appears in two sections

BWS best-worst scaling, *CHU9D* Child Health Utility 9 Dimensions, *cTTO* composite time trade-Off, *EQ, DCE* discrete choice experiment, *eTTO* experienced time trade-off, *HUI2* Health Utility Index Mark 2, *HUI3* Health Utility Index Mark 3, *LOD* location of dead, *PC* paired comparison, *QALY* quality-adjusted life-year, *SD* standard deviation, *SG* standard gamble, *T1D* type 1 diabetes, *TTO* time trade-off, *VAS* visual analogue scale, *WTP* willingness to pay

△ Adis

88], one from China [44], one from Japan [53], three from the Netherlands [30, 50, 61], one from New Zealand [9], one from Slovenia [60], five from the UK [8, 29, 89–91], seven from the USA [11, 51, 92–96] and two multi-country studies [15, 97]. Elicitation techniques used in studies in the group included TTO [8, 9, 44, 48, 53, 60, 91–93], (note that in some studies TTO was used to anchor the DCEs or BWS and not for comparative purposes), SG, [12, 29, 33, 88–90, 94, 95], VAS [9, 12, 30, 61, 89, 91] WTP [92, 93], BWS [44, 45, 48] and DCEs [11, 15, 50, 53, 60, 87, 96, 97] (some studies used more than one technique). Table S5 of the ESM contains study, country, title, aims, condition, sample size and age, perspective and information on values.

Standard gamble was used extensively in earlier papers (1996–2005 [12, 33, 88–90, 94]) and only once after 2005 (in 2019) [95]. Time trade-off has been used regularly from 2005 to 2021 [8, 9, 11, 44, 47, 53, 60, 92, 93]. Visual analogue scale was also used regularly over time [9, 12, 30, 61, 89, 91]. Best-worst scaling was used in 2012 [45], 2016 [47] and 2019 [44]. Discrete choice experiments were used from 2016 onwards [15, 50, 51, 53, 60, 87, 96, 97]. Health states being valued were variable, with 14 (52%) being generic and the remainder covering a range of acute, chronic and behavioural conditions. Similarly, where adult preferences were being elicited, perspectives varied from wide age groups to a specific age and values included child, parents and adult representatives of the general public, with no clear patterns between the values obtained from different perspectives. Reported values or utility decrements (when possible) are included in Table 3 as these reflect variations in perspective, values and elicitation methods (comparison of values between conditions is not meaningful here).

4 Discussion

The valuation of child health is key to conducting economic evaluations to inform decisions on the reimbursement and pricing of health interventions for children. Previous reviews have investigated the measurement of child PROMs [5, 19]. In the current review, we have focused on valuation approaches. Whilst the processes of valuing health in adults are generally well established, there is little agreement on how children's health should be valued. Despite there being several generic PROMS used for the valuation of child health interventions (such as the EQ-5D-Y and the CHU9D), this review shows that there are a range of fundamental uncertainties as to how the health states described by these instruments should be valued. These uncertainties include appropriate methods for preference elicitation, and the perspective and sources of values that should underpin the valuation of child health. The different approaches that have been used have led to diverse findings across methods.

Our review has shown a growing trend in the use of elicitation methods such as DCEs and BWS surveys, and this trend has also been noted in the adult literature [98]. The use of methods such as SG and TTO when valuing child health has been relatively consistent over time, with the use of SG decreasing since the mid-2000s. The current use of TTO is predominantly limited to anchoring DCE and BWS preferences to a utility scale (see Shah et al. [66]). There have, however, been a few studies that have undertaken comparative evaluations of elicitation methods (though we note that observing differences in outcomes between methods may not explain how consistent and valid these methods are). None of the 30 studies that compared elicitation methods included comparisons between valuations obtained from DCE/BWS-based methods and TTO/SG-based elicitation methods. The increasing use of DCE/BWS-type elicitation methods noted in our review may reflect a perception that these methods might be less challenging for children and adults than TTO and SG methods. Obtaining consent from ethics committees for DCE/BWS studies may be less problematic as there are no life/death trade-offs such as in TTO and SG. Furthermore, some methods may vary in applicability dependent on the complexity of the descriptive system. For instance, in a study on the CHU9D [49], researchers concluded that BWS was easier to manage than DCEs for larger descriptive systems, whereas for a more concise descriptive system such as the EQ-5D-Y, a DCE may be easier to present and complete. Given the heterogeneity of methods used between studies as well as the differing perspectives and values, a comparison of values between methods was not meaningful.

A wide range of perspectives (whose health state the participant is being asked to value) have been used. These include valuing one's own health, others' health including one's own child or a hypothetical child of varying age. Likewise, there was variation in who was asked to value the health state including adults, adolescents and children representative of the general population or from selected groups. An example of the influence of perspective on values is shown in the study by Tejwani et al. [83] where utilities were lowest when caregivers made TTOs from their own lives, intermediate when time was traded from both the caregiver's and child's life, and highest when traded exclusively from the child's life.

There was also evidence of the influence of condition on perspective from selected groups including children/adolescents with a particular condition, adults who had the condition as a child or parents of children where the condition was also relevant. Differences in utilities may be expected amongst those with and without a condition, whether from their own perspective or as a parent proxy given the different level of knowledge and experience of the condition [99]. Whilst the studies identified in this review include health states for generic and a range of chronic, acute, fatal and

Table 3 Study, condition, perspective adopted (who is being asked about), source of values (who is completing task?), elicitation methods and measures and values (mean and/or median where information is presented in papers), by year, for Group 3

Author, year	Condition	Sample and sample age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers)
Torrance, 1996 [12]	Generic health states	Adults, parents of childhood cancer patients ($n = 59$) Parents from the general population ($n = 293$), latter only reported	Child aged 10 years who must live with condition until aged 70 years	Adults	VAS and SG	SG present limited number of health states Mobility 3/5: 0.78 ± 0.19 Fertility 3/3: 0.88 ± 0.14 Interior 1: 0.76 ± 0.23 Interior 3: 0.51 ± 0.29 VAS Present extended number of health states below are the comparisons with SG Mobility 3/5: 0.54 ± 0.24 Fertility 3/3: 0.61 ± 0.26 Interior 1: 0.34 ± 0.20 Interior 3: 0.21 ± 0.20
Juniper, 1997 [88]	Asthma	Children aged 7–17 years with asthma ($n = 52$)	Child, self	Children aged 7–17 years	SG	Correlations reported discussed but no values reported
Saigal, 2000 [33]	Extremely low birth weight	Parents of adolescents aged 12–16 years ($n = 275$)	Adolescent (aged 12–16 years) on four hypothetical health states	Parent	SG	Parents of ELBW teens mean (SD) = 0.91 (0.20); median = 1.0; range – 0.10 to 1.0 Parents of control teens mean (SD) = 0.97 (0.08); median = 1.0; range 0.45–1.0
McCabe, 2003 [29]	Generic health states	General public ($n = 198$)	Child aged 10 years, and expected to live for another 60 years	Parents of school-aged children	SG	Values ranged from: – 0.052 to – 0.248
Raat, 2004 [30]	Generic health states	Adults Parents of elementary school children ($n = 1920$)	Child aged 4–13 years	Adult	VAS	Mean scores for the best and worst HUI3 states were 97.05 (SD 4.67) and 6.08 (SD 8.96), $n = 1224$. Mean scores for the 63 other states were between these extremes ($n = 68$–86). The mean score for "Being dead" was 10.29 (SD 15.73; $n = 722$)

Table 3 (continued)

Author, year	Condition	Sample and sample age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers)
Lee, 2005 [93]	Pertussis-related health states for disease and vaccination	Adult patients (aged ≥ 18 years) and parents of adolescent patients (aged 11–17 years) with confirmed pertussis diagnosis ($n = 515$ total, $n = 303$ for TTO, $n = 309$ for contingent valuation)	Adults patients: self for prevention of infant health states (due to pertussis) in a 1-month old infant Parent: their adolescent child for prevention of infant health states (due to pertussis) in their child at 1 month of age	Adults	TTO, contingent valuation, i.e. WTP	Adult short-term health states: mean value range = 0.81–0.95 Adolescent short-term health states: mean value range = 0.67–0.92 Infant short-term health states: mean value range = 0.51–0.58 Adult long-term health states: mean value range = 0.92–0.995 Adolescent long-term health states: mean value range = 0.82–0.97 Infant long-term health states: mean value range = 0.77–0.82 Adult short-term health states: mean WTP range = $8–$8748 Adolescent short-term health states: mean WTP range = $18–$4265 Infant short-term health states: mean WTP range = $13,016–$19,426
Matza, 2005 [94]	Attention-deficit/hyperactivity disorder	Adult parents of children (age range not specified, mean age = 10.2 years) who were diagnosed with attention-deficit/hyperactivity disorder ($n = 43$)	Their child	Adult parent	SG	Mean value range (adjusted SG values) = 0.90–0.98
Secnik, 2005 [89]	Attention-deficit/hyperactivity disorder	Parents, average age 42.6 years ($n = 83$)	Own child (mean age of 12.6 years)	Parents	SG, VAS	Adjusted SG Range: 0.88–0.96 Child's own health state 0.91 (SD 0.13) Adjusted based on worst raw SG value of 0.66 (0.35) VAS range 87.1–30.2 Child's own health 56.6 (21.9) Worst health state 9.4 (9.9)
Stevens, 2005 [90]	Atopic dermatitis	Adults general population ($n = 137$)	10-year-old child	Adults	SG (gamble being full health and death)	Mean worst state 0.356 (SD 0.363) Mean best state 0.841 (SD 0.188)

Table 3 (continued)

Author, year	Condition	Sample and sample age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers)
Lee, 2010 [92]	Group A Strepto-coccus disease and vaccine adverse events	Parents of children (aged <18 years) diagnosed with Group A Streptococcus pharyngitis (n = 119)	Their child	Adult parent	TTO, WTP	Range of median days traded (undiscounted) = 0.42–90.0 days; Range of median days traded (discounted at 3% = 0.12–31.0 days; Range of median days traded (discounted at 5%) = 0.05–14.2 days; Range of median WTP = $30–$3000; Range of median WTP per QALY = ~$18,000/QALY–~$60 000/QALY
Moodie, 2010 [9]	Overweight and obese	Adolescents in a classroom setting (n = 279)	Self	Adolescents	TTO (using VAS, then recalibrated)	Values not reported
Summerfield, 2010 [91]	Deafness	Opportunistic sample (n = 180) of clinicians/research-ers, university students and parents (none of a child with a hearing deficit) Aged 19–71 years	Hypothetical own child	Adult	VAS and TTO Imagine they are 33 years with a profoundly deaf 6-year-old daughter. The TTO asks them how many years of their own life they would give (assuming they have 50 years to live) for the child to have normal hearing	TTO values reported as increments; Range in mean increments were 0.05 (2 implants vs one implement plus one hearing aid) 0.16 (1 implant plus hearing aid vs no implant); VAS values reported as increments; Range in mean increments were 0.06 (2 implants vs one implement plus one hearing aid) 0.25 (1 implant plus hearing aid vs no implant)
Beusterien, 2012 [8]	Hunter syndrome	Two samples: 1) Adults aged over 18 years (n = 311) 2) Parents (n = 27) and children with Hunter syn-drome aged 12–18 years (n = 11)	Child/adolescent	1) General population 2) Parents of and ado-lescents with Hunter syndrome	TTO (trading 10 years of remaining life)	Average values: Best health state 0.99 Worst health state 0.41
Ratcliffe, 2012 [45]	Generic health states	Adolescents aged 11–17 years (n = 590)	Self	Adolescents	BWS	Adolescent range: 0.329–0.963 after re-anchored using adult SG for PITS Adults values: 0.326–0.967; difference range: − 0.062 to 0.180
Craig, 2016 [51]	Child behavioural problems	Adults (n = 4155) aged 18 years or above	Child aged 7 or 10 years	Adult, general popula-tion	Paired comparison (type of DCE)	Data provided as QALYs and not easily extracted as values

Table 3 (continued)

Author, year	Condition	Sample and sample age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers)
Craig, 2016 [11]	Generic health states	Adults (n = 4155) aged 18 years or above	Child (age ranged between 7 and 12 years) with 10 years yet to live	Adult, general population	Paired comparison (type of DCE)	No utilities presented QALY loss: range of means = 0.151–2.004 QALYs
Craig, 2015 [96]	Generic health states	Adults (n = 4155) aged 18 years or above	Child aged 7 or 10 years	Adult, general population	Paired comparison (type of DCE)	Data provided as QALYs and not easily extracted as values
Ratcliffe, 2016 [48]	Generic health states	Adolescents 11–17 years (n = 1982)	Self	Adolescents	BWS TTO	4 chosen health states TTO – 0.2118 to 0.6263 BWS 0-1 scale: range 0–0.6027 Rescaling method 1: range – 0.2118 to 0.5186 Rescaling method 2: range – 0.1059 to 0.5606
Rowen, 2018 [50]	Generic health states	Adults representative of general population (n = 1276)	Self (as adult)	Adults	DCE	Range: – 0.568 (worst state) to 1 (best state). Anchored values not reported, only utility decrements
Chen, 2019 [44]	Generic health states	BWS: (n = 902) students aged 9–17 years, mean age of 13 years TTO: (n = 38) students mean age 18 years	Self	Children/adolescents and young adults	BWS and TTO	BWS was used to develop preference weights Equivalent to TTO health states before re-scaling were: worst: 0 highest: 0.71 TTO was used to rescale the BWS to a QALY scale based on 5 health states Worse state: – 0.086 (SD 0.42) Highest value for the health states: 0.73 (SD 0.42)
Jabrayilov, 2019 [97]	Generic health states	Adults from general population (n = 1409) in Hong Kong, USA and UK Adult caregivers of 0- to 3-year-olds from Hong Kong, USA and UK (n = 1229)	Child aged 0–3 years	Adults	DCE	DCE coefficients only reported, no values
McElderry, 2019 [95]	Cancer: responding to treatment, not responding, stable on treatment	Adults (n = 167) waiting for paediatric visits Excluded subjects who ever had a child with cancer	Child	Adult	SG	Values for all three "cancer" scenarios median 0.61 (IQR: 0.29–0.86), n = 88 Aggregate "serious illness" scenarios 'median of 0.72 (IQR: 0.42–0.92), n = 83

Table 3 (continued)

Author, year	Condition	Sample and sample age	Perspective adopted	Source of values	Elicitation methods and measures	Elicitation methods and reported values (as presented in papers)
Krabbe, 2020 [15]	Generic health states	Adults from general population (n = 1409) in China (Hong Kong), USA and UK Adult caregivers of 0- to 3-year-olds from China, USA and UK (n = 1229)	Child	Adult	DCE	Values range = 0.015–0.961 (health state 1111112 was reference level for interaction domain and represented perfect health, i.e. value = 1.000)
Retra, 2020 [61]	Generic health states	University students aged 18–50 years (n = 311)	Child aged 4, 10 and 16 years	Adults	VAS	Range of means = 39.2–74 Range of mean differences between ages = − 1.695 to 3.809
Bahrampour, 2021 [87]	Cerebral palsy	Adults (general public) aged over 18 years (n = 2002)	Child	Adults from the general public	DCE	Provides values for each level of each domain from which health state values can be calculated The decrements are broadly similar across all domains Highest decrements ranged from − 0.301 for pain/discomfort to − 0.218 for communication
Prevolnik Rupel, 2021 [60]	Generic health states	Adults aged >18 years representative of general community (n = 1276) DCE, n = 1074 TTO, n = 202	Child aged 10 years	Adult	DCE, TTO from sample to anchor	Mean cTTO scores ranged from − 0.691 for state 33333 to 1.000 for state 11111 (n = 202) DCE 0.265 (mean) ± 0.326 (SD); range: − 0.691 to 1 (n = 1074)
Shiroiwa, 2021 [53]	Generic health states	Adults aged 20–79 years (n = 1047)	Child aged 10 years	Adults	cTTO and DCE (not a comparison, used together)	TTO used to anchor DCE by mapping to the 26 health states valued: range 0.20–0.94

SD and other variance measures are included in the ESM

BWS best-worst scaling, *cTTO* composite time trade-off, *DCE* discrete choice experiment, *HUI3* Health Utility Index Mark 3, *IQR* interquartile range, *PC* paired comparison, *PITS* lowest level health state of the CHU9D, *QALY* quality-adjusted life-years, *SD* standard deviation, *SG* standard gamble, *TTO* time trade-off, *VAS* visual analogue scale, *WTP* willingness to pay

non-fatal conditions, it was not possible to discern the likely magnitude of the difference in utilities assigned by the general public and those with experience of the condition. Lloyd et al. [57] suggest that respondents' perceptions of HRQoL impacts were not only influenced by the physical effects of a condition, but by external factors as well, such as patients with type 1 diabetes familiarity with needling and therefore reduced disutility for infusion therapy when compared with the general public. The influence of condition has also been shown in other contexts. [100]

Our review identified 23 studies where a comparison between perspectives (i.e. whose health state is being valued, own, other) can be made and these tended to suggest that children provide lower utilities than adults for the same health states (Group 2 studies, see Table 2). However, there was little discussion by study authors of the implications of this finding. Because of the mix of perspectives and values, it was not possible to evaluate possible bias in outcomes or the viability of the various methods for valuing child health. Furthermore, the preference elicitation tasks in these studies was dominated by TTO, SG and VAS. This limits the assessment of possible interactions between methods, particularly given the increasing use of DCEs and BWS. We were also unable to make any conclusions regarding feasibility (dropout rates, time taken to complete survey, difficulty of task) because of the very limited information presented in the included studies.

When adults are asked to value child health states, length of survival seems to be viewed as more important relative to quality of life than when adults are asked to value adult health states. This can be an issue for the comparability of these values and their use in economic evaluations [101], and it raises the question of whose values should be sought when valuing children's health. An argument used in the valuation of adult health is that the adult general public, as taxpayers and potential beneficiaries from publicly funded healthcare, should be the source of valuations, and this argument is used by bodies such as the National Institute for Health and Care Excellence in England and Wales [102]. The same principle might be taken to suggest that the preferences of the adult general public should also determine values for child health states; however, children are also potential beneficiaries of healthcare services, and older children may contribute financially through the tax system. Generally, societies tend not view children as autonomous legal, social and economic agents. Legal distinctions are typically made between children and adults across a wide range of behaviours and responsibilities, for example in the areas of drinking, driving, voting and making contracts, with different age cut-offs often used for these different behaviours. However, given that older children (e.g., 16- to 17-year-olds) have been found to be able to provide reasonable responses to valuation exercises [78, 103], and, in many countries, a proportion of children in this age group have regular engagement in the workforce and may therefore also contribute to the public purse through income tax contributions, we raise the question about who should be considered the "general public", and whether adolescents should be included in valuations for adults' HRQoL? These normative issues impact on methods and need further exploration.

Another consideration arising from this review is that most of the PROMs available for measuring child HRQoL contain states that may or may not be considered to be worse than dead; however, whether and how the studies allowed for states worse than dead, and what the minimum value is that preference elicitation methods could in principle produce, was often absent or poorly described. Only ten of the 77 studies appeared to allow for values less than zero, with the majority anchoring values to 0 (death or worst health state relevant to non-fatal conditions) to 1 (full or best imaginable health). In only a few studies was it clearly stated how anchoring to dead was undertaken. As noted by Shah et al. [52], adults may value states that are perceived to be worse than dead (or very poor health states) differently for themselves than they do when acting as a proxy for children, reflecting an unwillingness to trade off length of life for children.

4.1 Strengths and Limitations

Interest from the research community, policy makers and practitioners in valuing child health states has grown rapidly in recent years and we are aware of several studies currently underway (particularly to value the EQ-5D-Y) and others currently being considered for publication that could be included in future reviews. Our review takes a snapshot of the current situation but this area is changing quickly, and the outcomes of these studies, and whether they provide any additional clarity to our research questions, will need to be tracked over time. The search strategy used was broadly based using subject headings (MESH) and text words aimed at identifying studies on quality of life in populations of children and young adults. Subject headings and text words were used to filter the broad search terms to identifying studies addressing methodological issues. Although the methods concepts and text words were broad, there remains a possibility that relevant published studies may have been missed if they were not indexed according to the subject headings and did not describe the methods used in the abstract and keywords that match the terms used in the methods filter. Papers were only included if published in English. We also note that we limited our data extraction to the information that was contained in the papers, and that no contact was made with authors.

Whilst our review has identified over 70 studies published since 1996 that address elicitation methods for the

valuation child health states, we were not able to draw clear conclusions on methodological issues because of the ad hoc nature of research in this field and inadequate reporting of key details of the studies. There are several key questions that require focused and well-designed research programmes to address. Fundamental aspects that remain unanswered include the influence on health state valuations of differing perspectives, values and elicitation methods as well as methods for anchoring scales (particularly relevant to DCEs and BWS). In respect of the latter two methods, the evidence does not support one method over another, and there are many issues still unresolved or unreported. We also found that measures of feasibility such as time to completion, participant dropout or qualitative approaches such as think-aloud assessments or interviews were not reported. When specifically addressed, feasibility was generally described as being demonstrated by achieving valid/sensible results with no feasibility testing. This shortcoming is a feature of research in this area more generally; for instance, in a recent systematic where out of 110 versions of 89 PROMs specifically developed for children, only two included feasibility testing [5]. As noted by Rowen et al. [1], some of these questions (e.g. perspectives and values) are essentially a matter of judgement (i.e. normative); however, judgements about differences between alternate normative positions need to be informed by evidence. A further question that requires more research is how to measure which elicitation method might work best and in which circumstances (such as ease, cognitive burden and evidence of feasibility), which could not be determined from the findings in this review. The authors acknowledge that there are challenges and issues related to evaluating valuation studies and that this can lead to philosophical differences between researchers. The authors have aimed to make these issues clear, and to come to reasonable conclusions given the limitations of the data.

A major challenge that emerged from this review was the significant variation in what information was reported in the papers, as well as the varying amount of detail provided on the methods used to generate utility values. There is a clear need for standards of reporting that reflect the specific requirements of child health valuations [104]. A recent review by Zoratti et al. [105] has highlighted the need for reporting standards for adult health valuation studies. However, as child health valuation presents a range of additional challenges, there is a strong need for child-specific reporting standards. Without such standards, it is not possible to summarise collective evidence to support normative positions or standardised requirements for health state valuations to support policy. Given the importance of understanding valuation for child HRQoL, guidelines for reporting of future studies would be likely to improve the overall quality of publications and enhance the comparability of research results.

5 Conclusions

This review summarises available evidence for a range of research questions relevant to valuations of child health, including whose values and perspectives are most relevant and how best to address the methodological challenges. The use of elicitation methods has changed over time, with recent studies favouring methods such as DCEs; however, the literature provides few meaningful data as to which methods are preferable for obtaining values for child HRQoL. Differences in reporting limited the conclusions that could be formed. Difficulties encountered in drawing conclusions from the data suggest that reporting guidelines are required to improve the consistency and quality of reporting of studies that value children's health using preference-based techniques.

Declarations

Conflict of interest The authors declare that they have no conflicts of interest. We note that ND, RV, DJS, RN and BM are members of the EuroQol Group and that ND, RV, DJS, RN, JR, BM, EL and CB have received funding through the EuroQol Group for related research. ND is the Chair of the Board of the EuroQol Research Foundation.

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Code availability Not applicable.

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