



Deposited via The University of Sheffield.

White Rose Research Online URL for this paper:

<https://eprints.whiterose.ac.uk/id/eprint/201425/>

Version: Accepted Version

Article:

Gale, V. and Carlton, J. (2023) Including young children in the development and testing of patient reported outcome (PRO) instruments: a scoping review of children's involvement and qualitative methods. *The Patient - Patient-Centered Outcomes Research*, 16 (5). pp. 425-456. ISSN: 1178-1653

<https://doi.org/10.1007/s40271-023-00637-8>

This version of the article has been accepted for publication, after peer review (when applicable) and is subject to Springer Nature's AM terms of use, but is not the Version of Record and does not reflect post-acceptance improvements, or any corrections. The Version of Record is available online at: <http://dx.doi.org/10.1007/s40271-023-00637-8>

Reuse

Items deposited in White Rose Research Online are protected by copyright, with all rights reserved unless indicated otherwise. They may be downloaded and/or printed for private study, or other acts as permitted by national copyright laws. The publisher or other rights holders may allow further reproduction and re-use of the full text version. This is indicated by the licence information on the White Rose Research Online record for the item.

Takedown

If you consider content in White Rose Research Online to be in breach of UK law, please notify us by emailing eprints@whiterose.ac.uk including the URL of the record and the reason for the withdrawal request.

1 **Title:** Including young children in the development and testing of patient reported outcome (PRO) instruments: a
2 scoping review of children’s involvement and qualitative methods

3

4 **Running heading:** Scoping review of young children’s involvement in qualitative research for PROM
5 development

6

7 **Author Information:**

8 Names – Victoria Gale (Orcid ID: 0000-0001-7472-2039) and Jill Carlton (Orcid ID: 0000-0002-9373-7663)

9 Affiliation – The University of Sheffield, School of Health and Related Research, Sheffield, UK

10 Corresponding author – Victoria Gale, vargale1@sheffield.ac.uk

11

12 **Statements and Declarations:**

13 Funding – This review is part of PhD research funded by SF-6D royalty income.

14 Conflicts of interest – Victoria Gale and Jill Carlton declare that they have no conflicts of interest.

15 Data availability statement – The datasets generated during and/or analysed during the current study are
16 available in Online Resource 4.

17 Author contributions – All authors contributed to study conception, design, and review preparation. Data
18 extraction and analysis was performed by VG. VG prepared the first draft of the manuscript and both authors
19 commented on previous versions. The final version was read and approved by both authors.

20 Acknowledgements – We would like to thank John Brazier for his support conceptualising the review and
21 methodology, and ongoing help throughout the review process. We would also like to thank Louise Flazon for
22 her support developing the search strategies for online databases.

23 Ethics approval and consent to participate and publication – Not applicable.

24

25

26 **Abstract**

1 **Background** Qualitative research during the development/testing of Patient Reported Outcome Measures
2 (PROMs) is recommended to support content validity. However, it is unclear if and how young children (≤ 7 years)
3 can be involved in this research because of their unique cognitive needs.

4 **Objectives** To investigate the involvement of children (≤ 7 years) in qualitative research for PROM
5 development/testing. This review aimed to identify (1) which stages of qualitative PROM development children
6 ≤ 7 years had been involved in, (2) which subjective health concepts had been explored within qualitative PROM
7 development with this age group, and (3) which qualitative methods had been reported and how these compared
8 existing methodological recommendations.

9 **Methods** This scoping review systematically searched three electronic databases (searches re-run prior to final
10 analysis on 29th June 2022) with no date restrictions. Included studies had samples of at least 75% aged ≤ 7 years
11 or reported distinct qualitative methods for children ≤ 7 years in primary qualitative research to support concept
12 elicitation or PROM development/testing. Articles not in English and PROMs that did not enable children ≤ 7
13 years to self-report were excluded. Data on study type, subjective health, and qualitative methods were extracted
14 and synthesised descriptively. Methods were compared to recommendations from guidance.

15 **Results** Of 19 included studies, 15 reported concept elicitation research and four reported cognitive interviewing.
16 Most explored quality of life (QoL)/health-related quality of life (HRQoL). Some concept elicitation studies
17 reported creative/participatory activities had supported children's engagement, but results and reporting detail
18 varied considerably across studies. Cognitive interviewing studies reported less methodological detail and fewer
19 methods adapted for young children compared to concept elicitation studies. They were limited in scope regarding
20 assessments of content validity, mostly focussing on clarity while relevance and comprehensiveness were
21 explored less.

22 **Discussion** Creative/participatory activities may be beneficial in concept elicitation research with children ≤ 7
23 years, but future research needs to explore what contributes to the success of young children's involvement and
24 how researchers can adopt flexible methods. Cognitive interviews with young children are limited in frequency,
25 scope, and reported methodological detail, potentially impacting PROM content validity for this age group.
26 Without detailed reporting, it is not possible to determine the feasibility and usefulness of children's (≤ 7 years)
27 involvement in qualitative research to support PROM development and assessment.

28 **Key Points for Decision Makers**

- 1 • Qualitative research is widely recommended to be used during the development/testing of Patient Reported
2 Outcome Measures (PROMs) to establish instrument content validity.
- 3 • There are few examples of children aged ≤ 7 years being involved in cognitive interviews to test PROMs and
4 in the absence of detailed reporting it is difficult to establish if or how the challenges of involving this
5 population in cognitive interviews can be overcome.
- 6 • Children ≤ 7 years have had more involvement in concept elicitation research but reporting of methodological
7 adaptations to meet their unique needs is not well-established.

8

9

10

11

12

13

14

15

16

17

18

19

20

21

22

23 **Including young children in the development and testing of patient reported outcome (PRO) instruments:**
24 **a scoping review of children's involvement and qualitative methods**

1

2 **1 Introduction**

3 Children's self-reports of health have become increasingly valued in health care and research [1-3] and Patient
4 Reported Outcome Measures (PROMs) are one way they can be measured. PROMs are standardised health
5 questionnaires that measure an individual's health from their own perspective without interpretation by others [4,
6 5]. They do not assess biomedical markers (e.g., blood pressure), but instead provide quantitative descriptions of
7 patients' subjective health perceptions and outcomes, such as symptoms and functioning [3, 6, 7]. Quality of life
8 (QoL) and health-related quality of life (HRQoL) are also commonly measured with PROMs [8]. However, the
9 terms QoL and HRQoL can result in confusion in the literature; they are notoriously difficult to define resulting
10 in lack of consensus over meaning, and the terms are often used interchangeably even though they are different
11 [8]. PROMs can be specific to a health condition (such as diabetes) or generic (designed to measure health across
12 multiple conditions) [7, 9, 10].

13 By directly measuring aspects of health considered to be known best by the patient themselves (e.g., symptoms
14 and functioning), children's PROMs can help ensure their health care is patient-centred [3-11]. PROMs have a
15 range of applications, such as in health care to support patient assessment, diagnosis, and monitoring, in clinical
16 trials to support the evaluation of new treatments, and in the evaluation of health care systems [3, 4, 6, 12, 13].
17 Paediatric PROMs can also be completed by proxy, such as by a parent on behalf of their child. However, this
18 inevitably involves inference from the proxy and the literature reports mixed results regarding the level of
19 agreement between children's self-report and proxy report [3]. As such, it is recommended by the FDA that
20 children self-report using PROMs wherever possible [4]. This review focusses specifically on young children's
21 self-report.

22 It is essential that the content validity of PROMs is established to ensure accuracy and appropriateness for the
23 target population [4, 14-16]. Content validity is the extent to which the PROM contains health concepts that are
24 important and comprehensive for the target population and can be established through the development and testing
25 process [3, 4, 16, 17]. PROM development typically proceeds through four main stages: (1) concept definition
26 i.e., creating a conceptual framework that defines and describes the health concept to be measured; (2) content
27 generation i.e., developing the draft PROM; (3) cognitive testing and refinement i.e., assessing the clarity and
28 comprehensiveness of the draft PROM among a sample of the intended target population and modifying the
29 PROM as needed; and (4) psychometric testing i.e., assessing the validity and reliability of the PROM through

1 quantitative methods (Figure 1, see [13] for a detailed description of this). Qualitative research with
2 representatives of the target population is widely recommended as good practice during stages one and three to
3 establish and provide evidence for content validity [4, 14, 15, 17, 18]. This can include concept elicitation and/or
4 cognitive interviewing.

5 Concept elicitation studies consist of interviews and/or focus groups where representatives of the target population
6 are asked about their experiences and perceptions of health. The results are then used to develop a conceptual
7 framework which helps identify and define the health concepts to be included in the PROM [4, 15, 17-19].
8 Cognitive interview studies are used to test and refine PROMs [e.g., 4, 17]. This specific method of interviewing
9 aims to evaluate whether questionnaires are clear, relevant, and comprehensive for the intended target population.
10 The interviews could involve participants being asked direct questions about a PROM ('verbal probes') or asking
11 participants to say all their thoughts out loud ('think aloud') whilst they complete a PROM [14, 17, 20]. Content
12 validity of an existing PROM developed previously can be tested through cognitive interviews and concept
13 elicitation interviews [21]; a conceptual framework developed from concept elicitation interviews can be
14 compared to the content of a pre-existing PROM.

15 However, while it is recommended to involve children in both concept elicitation research and cognitive
16 interviews wherever possible [3, 4, 22], this is not without its challenges. Children's developing cognitive skills
17 may result in limited understanding of the qualitative task or of their role within research, they may have
18 difficulties understanding the health concept of interest, and power imbalances between researchers and children
19 may cause children to feel obliged to respond or to adjust responses to 'please' adults [3, 22]. These challenges
20 are likely to be greater for younger children (i.e., ≤ 7 years) because they are at an earlier stage of development;
21 for example, they are more susceptible to social desirability bias and will struggle more with abstract questions
22 [13]. Involving children in qualitative research is further complicated by the dynamic nature of development where
23 chronological age does not necessarily correspond to cognitive ability [13].

24 Guidance exists for how to adapt qualitative methods to mitigate these challenges [3, 13, 22], however, this is
25 limited for children aged ≤ 7 years. Furthermore, few examples of children aged ≤ 7 years being involved in
26 qualitative research for PROM development/testing have been identified in the literature [10, 23-26] and in
27 focussing on children aged up to 18 years, previous reviews have not isolated the level of involvement of children
28 ≤ 7 years specifically [10, 23-26]. A recent review by Husbands et al [24] explored children's (0-18 years)
29 involvement in concept elicitation research and found few examples of methods having been adapted for the needs

1 of children, such as utilising creative and/or participatory activities. They also found a general lack of transparency
2 in reporting, particularly for data analysis and sampling [24]. The current review builds on these findings by
3 focussing specifically on the involvement of children ≤ 7 years and by exploring a broader range of qualitative
4 research for PROM development/testing (i.e., cognitive interviewing, content validity testing). It also compares
5 reported qualitative methods to recommendations from guidance.

6 The known challenges of involving young children in qualitative research, limited guidance, few examples in the
7 literature, and broad focus of previous reviews on childhood (i.e., 0-18 years) means it is unclear whether it is
8 possible to enable children ≤ 7 years to participate meaningfully in qualitative research for PROM
9 development/testing. This scoping review aims to investigate the involvement of children aged ≤ 7 years in
10 qualitative research for PROM development/testing¹. It asks the following questions:

- 11 1. What stages of PROM development/testing (concept elicitation, content generation, cognitive interview,
12 content validity studies) are children aged ≤ 7 years being involved in through qualitative methods?
- 13 2. What subjective health concepts (e.g., symptoms, QoL) are being explored with children ≤ 7 years in
14 qualitative research for PROM development/testing or concept elicitation?
- 15 3. What adapted qualitative methods (as suggested by guidance [3, 13, 22]) are being reported in qualitative
16 studies for PROM development/testing or concept elicitation with children aged ≤ 7 years?

17

18 **2 Methods**

19 The review protocol was registered on the International Prospective Register of Systematic Reviews
20 (PROSPERO) before initial searches were conducted (registration no: CRD42022308007) and can be accessed at
21 https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=308007. Reporting follows the Preferred
22 Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) guidelines for scoping reviews (the
23 completed PRISMA-ScR checklist can be found in Online Resource 1).

24

25 2.1 Search strategy

26 With the support of an Information Specialist experienced in conducting health systematic reviews, a search
27 strategy was developed to retrieve a broad range of studies involving children ≤ 7 years in qualitative research for
28 PROM development and/or testing. It also sought to retrieve concept elicitation studies that aimed to develop a

1 conceptual framework for a subjective health concept without explicitly stating intentions to develop a PROM.
2 These studies were included given development of a conceptual framework is a key stage in PROM development
3 (see Introduction and Figure 1). The search was not limited to any specific health condition and could include
4 physical and/or mental conditions. For this review ‘subjective health’ was defined as any self-report coming
5 directly from the child of their experiences of health conditions and/or treatment (e.g., symptoms, pain) or their
6 perceptions of the impact of the health/treatment on their daily life (e.g., cognitive, emotional, or social
7 implications). This could include physical, mental, and/or social domains, in-line with the World Health
8 Organisation definition of health [27]. It did not include, for example, children’s reports of experiences in hospital.
9 Medline via Ovid was used to develop search terms combining four key concepts (Table 1). The search strategy
10 was then adapted for Embase and CINAHL (see Online Resource 2 for full search strategies for each database).
11 Databases were searched without date limits. Forward citation searches were conducted using the “find citing
12 articles” feature of Medline and PubMed. Reference lists of included studies and of four pre-existing systematic
13 reviews [10, 23-25] were searched by hand.

14

15 2.2 Identifying studies for inclusion

16 Studies were eligible for inclusion if (1) they were qualitative studies of concept elicitation, content generation,
17 cognitive testing, or content validity testing; (2) at least 75% of the sample were aged ≤ 7 years or distinct methods
18 were described for children ≤ 7 years; (3) subjective health had been explored with children ≤ 7 years; and (4)
19 where a PROM had been developed this enabled children ≤ 7 years to self-report. Articles not written in English,
20 reviews, conference abstracts, research protocols, and dissertations/theses were excluded, as well as studies that
21 did not report sufficient detail of qualitative methods to be useful in data analysis (e.g., a brief comment that
22 qualitative methods were used).

23 All references were imported to Endnote where duplicates were removed. Using Microsoft Excel, the first
24 reviewer (VG) screened all abstracts and full texts. The second reviewer (JC) conducted an independent check of
25 10% of the abstracts and 10% of the full texts and Cohen’s Kappa was used to calculate interrater reliability. The
26 reviewers met after each screening stage to discuss disagreements until consensus was reached. If there was not
27 enough information to determine whether the full inclusion criteria was met during the title and abstract stage, the
28 full text was retrieved for screening. A decision tree was used by both reviewers to guide full text screening

1 (Online Resource 2). Screening of forward citation searches of included studies and reference list searches of
2 included studies and existing reviews [10, 23-25] was conducted in the same way by the first reviewer only.

3

4 2.3 Data extraction

5 The first reviewer extracted data from included studies using data extraction forms in Microsoft Excel (Online
6 Resource 3). These included study details (e.g., aim, sample, element of PROM development, subjective health
7 concept), methodological details, and PROM details where developed (e.g., domains, recall period, response
8 options.). Where information could not be found about PROMs, attempts were made to contact authors. Missing
9 information was listed as “not reported”.

10

11 2.4 Synthesis methods

12 All data charting and synthesis was conducted by the first reviewer. As a scoping review, synthesis aimed to
13 provide a description of the included studies guided by the three research questions. Extracted data were tabulated
14 in Microsoft Word to support data charting and synthesis. First, included studies were organised by type (i.e.,
15 concept elicitation, cognitive interviewing). Next, subjective health concepts included in these studies were
16 identified through examination of author definitions of concepts and of the questions children were asked in the
17 qualitative research. Prior to the review being conducted, three guidance documents for conducting qualitative
18 research with children to support PROM development/testing [3, 13, 22] had been identified from initial scoping
19 searches and common methodological recommendations and considerations extracted (Table 2). These
20 recommendations broadly fell into four categories: (1) topic/method guide considerations; (2) practical
21 considerations; (3) additional/modified activities; and (4) methodological considerations specific to cognitive
22 interviewing (Table 2). These recommendations were used to guide synthesis for the third research question - data
23 were charted according to which recommendations had been reported by included studies and synthesis included
24 consideration of similarities and differences between studies. Author reflections on the success (or
25 unsuccessfulness) of involving young children in this type of research were also considered during analysis.

26 In addition, data charting and synthesis for the cognitive interviewing studies was guided through a comparison
27 of extracted data to recommendations from The International Society of Pharmoeconomics Research (ISPOR)
28 [14] and the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) [21].

1 These documents are used to support cognitive interviews during PROM development and recommend various
2 aspects of content validity (e.g., clarity, comprehensiveness) and aspects of PROMs (e.g., items, instructions,
3 response options) to be assessed during cognitive interviews. Recommendations from these documents are listed
4 in Table 3.

5

6

7 **3 Results**

8 3.1 Search results

9 Initial database searches were run on 1st February 2022 and re-run on 29th June 2022 (searched from February
10 2022 – June 2022). A final total of 19 studies were included (Figure 2). One study [29] was an exploratory
11 component of a larger study [30]. For the 10% check of screened abstracts, Cohen’s Kappa was 0.55 (indicating
12 moderate agreement, [31]) and percentage agreement between reviewers was 93%. The first reviewer was more
13 sensitive to including studies to avoid missing important information, possibly explaining the lower Kappa score.
14 Values for the 10% check of full texts did not meet the requirements for the Kappa statistic so percentage
15 agreement was calculated as an alternative. The reviewers independently agreed on 95.7% of the full texts (44/46).
16 Initial disagreements concerned whether methods for children ≤ 7 years were distinct and whether subjective health
17 was explored with children; after discussion, a consensus was reached to include both studies.

18

19 3.2 Study characteristics

20 Of the 19 included studies, 15 reported concept elicitation research and four reported cognitive interviewing
21 (Table 4). None explored generic health in a general population; eighteen were condition-specific (e.g., amblyopia
22 [32, 33], dysphonia [34], and cancer [29, 30] and one was domain-specific (exploring psychological functioning
23 in a sample of children without health conditions [35] (Table 4). Study aims broadly fell into two categories – to
24 conduct qualitative research to inform the development of a new PROM, or to conduct qualitative research to
25 explore the impact of a health condition on the lives of children affected, or a combination of both (Table 4).

26 Seventeen studies explicitly stated intentions to develop a PROM. Of the two remaining studies, one reported
27 developing a conceptual framework as an exploratory component of a larger PROM development study [29] and

1 one aimed to only develop a conceptual framework [36]. Sixteen unique PROMs were identified (PROM
2 characteristics in Online Resource 4). Most of the identified PROMs also had a proxy version available, and only
3 two were developed specifically for children ≤ 7 years (Child Amblyopia Treatment Questionnaire (CAT-QoL)
4 [32, 33]; Mini-SSPedi [30]).

5 When considering the youngest age of children included in studies, the youngest age of most study samples was
6 five years (Figure 3). Five studies had involved children younger than five years; three reported that children this
7 age were able to successfully participate in the research by engaging in the task and providing meaningful data
8 for use in analysis [29, 30, 32] while the remaining two reported that children this age had not been able to engage
9 or provide meaningful data [34, 37]. Sixteen studies also included children older than seven years but reported
10 distinct methods for children ≤ 7 years, hence meeting the inclusion criteria.

11

12 3.3 Subjective health concepts

13 Four categories of subjective health were identified: QoL/HRQoL (13/19 studies), symptoms (3/19 studies),
14 psychological functioning (2/19 studies), and participation (1/19 studies) (Figure 3 4). Author definitions of
15 QoL/HRQoL varied between studies, but all explored children's perceptions of the impact/effects/experiences of
16 a health condition on their lives in multiple situations e.g., home, school. Symptom studies investigated children's
17 reports of subjective indicators of a health condition (e.g., tiredness, nausea), psychological functioning studies
18 explored children's reports of their emotions and emotional distress (e.g., anxiety, anger), and the participation
19 study explored children's reports of how they attended and felt involved in activities.

20

21 3.4 Qualitative methods and comparison to methodological guidance

22 3.4.1 Concept elicitation research

23 Most concept elicitation research used individual interviews (11/15 studies). The remaining four studies described
24 focus groups, one of which offered optional individual interviews if children preferred [35]. Table 5 details
25 methods reported by concept elicitation studies; levels of reporting detail varied with nine studies providing
26 limited detail regarding children ≤ 7 years, [37-44], five of which were PedsQL module development studies [38,
27 39, 41-43]. Nine studies explicitly stated that young children's participation had been successful because they
28 engaged in activities and/or had been able to provide meaningful data [29, 32, 34-36, 39, 40, 45, 46]. Level of

1 reporting detail varied across these studies (Table 5) and four reported successful involvement was possible for
2 some, but not *all* children; Carlton [32] found five children were unresponsive in the interview; Connor et al [34]
3 found meaningful data could be gathered from children aged 5-7 years but not from children 2-4-years-old; Hyslop
4 et al [29] reported that drawing was only successful for 18/30 children; and Morris et al [45] reflected that the
5 individual maturity of the child determined the success of their engagement. Among the remaining six studies not
6 explicitly reporting success, one reported that young children's involvement had been unsuccessful [37] (it was
7 not reported why), and success was not discussed by the remaining five [38, 41-44].

8 Several methodological recommendations from the three guidance documents [3, 13, 22] were reported among
9 the nine studies describing successful involvement. Five reported adapted activities and authors explicitly
10 discussed these as being beneficial in enabling children's participation [29, 35, 36, 45, 46]. These activities
11 included photo elicitation and an adapted informed consent procedure [36], drawing [29], picture-card fishing
12 game [46], statement agreement and life-mapping [45], and multi-sensory response options [35] (Table 5).
13 Conversely, one study reported that the adapted activity (drawing) was distracting to children and so was
14 abandoned after several interviews [32]. Some practical considerations recommended by the guidance documents
15 [3, 13, 22] were also reported by these studies; six reported building rapport with children, [29, 32, 34-36, 45],
16 four reported ensuring children were comfortable in the setting [35, 36, 45, 46], and two reported recruiting focus
17 groups made up of siblings and/or peers such that children felt at ease and discussion was promoted [35, 46].

18 Other methodological recommendations from guidance [3, 13, 22] reported across concept elicitation studies
19 included facilitator training, consideration for the presentation of questions, and appropriate facilitator
20 communication (Table 6). The role of parents varied across concept elicitation studies; three describe parents as
21 being present but not involved [32, 35, 39], three as being present and supportive of their child's emotional needs
22 and communication [34, 36, 44], and two as taking part in a separate focus group to their children [40, 45]. Four
23 of the five PedsQL studies report parents were present but do not define their role in the interviews [38, 41-43],
24 and one report stated only that parents were not involved wherever possible [37]. Methodological
25 recommendations not regularly reported by concept elicitation studies included pilot testing, adapting the topic
26 guide across sessions, explaining recording equipment to children, and assessing children's understanding of the
27 task (Table 6).

28

29 3.4.2 Cognitive interviews

1 All four cognitive interviewing studies describe using individual interviews (Table 7). Two [30, 33] explicitly
2 stated that young children's involvement in cognitive interviewing was successful. Only one reported using an
3 adapted activity to enable children's involvement [30] (details in Table 7) which the authors discuss as being
4 necessary to meet the cognitive needs of the young children. It is unclear from the remaining three studies if
5 cognitive interviewing techniques were adapted to meet the needs of young children: two report using verbal
6 probes [47, 48] and one verbal probes and observation [33]. Two recommendations from guidance documents [3,
7 13, 22] were reported by all four cognitive interviewing studies - to follow the same general cognitive interviewing
8 procedure as is recommended for adults and to conduct the cognitive interviews within narrow age bands (Table
9 6). Aside from Tomlinson et al [30] very few other recommendations were reported by the cognitive interviewing
10 studies (Table 6).

11 Table 8 details which aspects of PROMs and which components of content validity (as identified from ISPOR
12 and COSMIN recommendations) were assessed by the cognitive interviewing studies. All four studies reported
13 assessing the clarity of items. Relevance was only assessed by two studies [47, 48] and comprehensiveness by
14 one [33] (Table 8).

15

16

17 **4 Discussion**

18 Nineteen studies were identified that reported involving children aged ≤ 7 years in qualitative PROM
19 development/testing or concept elicitation research, all of which were condition- or domain-specific. This could
20 reflect more condition-specific PROMs having been developed [49] or possibly that it is easier to conduct this
21 research where children have lived experiences of health conditions [50]. Five years was the most common
22 youngest age of samples. Similarly, guidance suggests children can be interviewed about their health from 5-8
23 years [3, 13, 22] when it is thought they would typically have developed a basic, concrete concept of health [19,
24 50]. However, this review found some evidence for the successful involvement of children younger than five years
25 (three studies reported children this age could engage in the research activities and provide meaningful data for
26 analysis [29, 30, 32]), in-line with guidance that age bands should be flexible because chronological age alone
27 does not determine ability [3, 13].

28

1 4.1 Concept elicitation studies

2 Successful participation for children ≤ 7 years was reported by more than half of the included concept elicitation
3 studies, suggesting it may be possible to involve this age group in concept elicitation research. Furthermore, the
4 included concept elicitation studies reported a variety of adapted methods (such as creative and participatory
5 activities), and several authors explicitly reported that these methods had been beneficial to enabling children's
6 (≤ 7 years) successful participation. Adapted methods may therefore be useful in enabling young children's
7 participation, as also concluded by Husbands et al [24] and existing literature within PROM development and
8 within qualitative research with children more broadly [3, 13, 22, 51].

9 However, this review has highlighted inconsistencies in the success of adapted methods and in the level of
10 reporting detail. Many studies did not report on the success of young children's involvement or did not provide
11 enough detail to determine what may have contributed to successful/unsuccessful involvement. Some authors
12 reported that adapted activities either did not work [37] or only worked for some children [29, 32, 34, 45] and did
13 not expand on whether alternative approaches were used to address this. Detailed methodological reporting is
14 necessary to ensure both trustworthiness in qualitative research [52], and to support methodological evaluation
15 when research methods are not well-established [53, 54]. This is also true of involving young children in concept
16 elicitation research. In the absence of detailed reporting across all included concept elicitation studies and variation
17 in approaches used by researchers, specific recommendations of methods cannot be made beyond a general
18 suggestion that adapted activities may be beneficial.

19 Future research should not only explore the potential benefits of using adapted activities with young children, but
20 also the factors that contribute to the success of these. Many variables likely contribute to the success of young
21 children's involvement, including their cognitive and communicative abilities and their understanding of the
22 health concept being discussed [55, 56]. Researchers involving young children in concept elicitation studies will
23 need to balance these needs, and because the current review has shown methodological approaches are not always
24 successful for every child, inevitably methods will need to be flexible. Therefore, while there is some evidence to
25 suggest it is possible to involve children ≤ 7 years in concept elicitation research, how this can be achieved
26 consistently remains unclear.

27

28 4.2 Cognitive interviewing studies

1 Only four cognitive interviewing studies were identified in this review, similar to previous reviews where very
2 few examples of cognitive interviewing studies with young children have been identified [10, 23, 25]. It is unclear
3 why there are few examples of cognitive interviewing with young children; in the absence of detailed reporting,
4 it is unknown whether unsuccessful attempts are being made and not reported, or not attempted at all. It is possible
5 that the unique features of cognitive interviews (i.e., answering questions *about* questions) are too challenging for
6 young children. This review suggests children from five years may be able to successfully engage in concept
7 elicitation research, while abstract cognitive interview questions are considered cognitively challenging, even for
8 adults [20, 57]. Young children are more likely to struggle with abstract questions, lengthy interview schedules,
9 and with providing explanations of their thinking [13, 22].

10 Alternatively, or perhaps in addition, researchers may be unsure of how best to adapt methods to enable young
11 children to take part. Young children's cognitive and communicative skills are still developing, and consequently
12 adapted methods of interviewing are needed to support them in taking part in cognitive interviews. This can
13 include ensuring children fully understand their role in the interview, allowing them longer to think about and
14 respond to interview questions, and adapting the language of the interview questions [3, 13, 22]. However, only
15 one study identified in this review reported adapting cognitive interviewing methods to meet the needs of young
16 children [30]. The remaining three provided limited detail of methods and reported few methodological
17 recommendations from guidance. This contrasts the concept elicitation studies where generally more adapted
18 methods and recommendations from guidance were reported. Limited reporting does not mean recommendations
19 were not implemented in the research but as discussed earlier, makes methodological evaluation challenging [54].
20 These findings may indicate that less is known about how best young children can be involved in cognitive
21 interviews compared to concept elicitation research. It could also be that the typically more structured procedure
22 of cognitive interviews may make it more challenging for researchers to implement flexible, creative approaches
23 adapted for the needs of young children.

24 The cognitive interviewing studies identified in this review were also limited in the extent to which they assessed
25 PROM elements and aspects of content validity recommended to be assessed by current best practice guidelines
26 [14, 21]. They primarily focussed on assessing clarity, while assessments of comprehensiveness, relevance, recall
27 period, response options, and instructions were less reported. Clarity may be easiest to assess with young children;
28 both direct questioning and observation could be used (e.g., [33]). By contrast, young children may struggle with
29 concepts of time if asked about recall period [13], their limited attention spans may make it difficult to conduct a

1 'full' cognitive interview [13], and comprehensiveness may be too abstract. One study sought to limit cognitive
2 load for young children, but still did not assess comprehensiveness [30].

3 Overall, the limitations identified in this review regarding cognitive interviews with young children could have
4 implications for the content validity and appropriateness of PROMs for this age group. Cognitive interviews are
5 recognised as being fundamental to establishing content validity [4, 14] without which, PROMs may collect
6 inaccurate information. As with concept elicitation research, more detail in methodological reporting is necessary
7 within cognitive interviews with young children. Furthermore, specific research is needed to explore how, or
8 indeed if, young children can take part in cognitive interviews to support PROM development testing. This is of
9 particular importance given the unique nature and aims of cognitive interviewing which present considerable
10 challenges when seeking to involve children ≤ 7 years.

11 This review is not without limitations. The exclusion criteria used meant several studies involving children < 7
12 years were excluded, as the methods and results were not distinct from those used with older children (i.e., > 7
13 years). As such, information about the involvement of children ≤ 7 years may have been missed. The number of
14 included studies, particularly cognitive interviewing studies, was small, which has made it difficult to draw
15 conclusions. In addition, a formal assessment of the methodological quality of the included studies has not been
16 conducted. However, formal quality assessments are not always typical within scoping reviews where the aim is
17 to provide a description of the current evidence base [58, 59]. Furthermore, in comparing methods of included
18 studies with current methodological recommendations, this review has identified a need for more detailed and
19 consistent reporting of methods from qualitative research with children in this area; this is necessary before
20 methodological evaluations can be made [53].

21

22 **Conclusion**

23 In conclusion this scoping review has identified substantial gaps within the reporting of qualitative concept
24 elicitation and cognitive interviewing studies with children ≤ 7 years. Creative/participatory activities may support
25 young children's involvement in concept elicitation research, but inconsistent reporting means it is unclear how
26 children's needs can be balanced with research aims to enable successful participation. Children ≤ 7 years have
27 been less involved in cognitive interviewing, and limited reporting makes it difficult to determine whether this is
28 because cognitive interviews are too challenging, or whether methods have not been adequately adapted.
29 Establishing content validity of PROMs is essential. As such, qualitative research aiming to develop or assess

1 content validity needs to be well-designed and described. In the absence of detailed reporting, replication and
2 evaluation of such qualitative methods with children ≤ 7 years is difficult, which has implications for the content
3 validity of PROMs developed for this age group. Consistent, comprehensive documentation is needed within this
4 area of research to be able to establish the feasibility and usefulness of young children's involvement in the
5 development and assessment of PROMs.

6

7

1 **References**

- 2 1. Cremeens J, Eiser C, Blades M. Characteristics of health-related self-report measures for children aged three to
3 eight years: a review of the literature. *Qual Life Res.* 2006; <https://doi.org/10.1007/s11136-005-4184-x>
- 4 2. Germain N, Aballéa S, Toumi M. Measuring health-related quality of life in young children: how far have we
5 come? *J Journal of Market Access & Health Policy.* 2019; <https://doi.org/10.1080/20016689.2019.1618661>
- 6 3. Matza, L.S., et al., Pediatric patient-reported outcome instruments for research to support medical product
7 labeling: report of the ISPOR PRO good research practices for the assessment of children and adolescents task
8 force. *Value Health.* 2013; <https://doi.org/10.1016/j.jval.2013.04.004>
- 9 4. (FDA), U.S.D.o.H.a.H.S.F.a.D.A. Patient-reported outcome measures: use in medical product development to
10 support labeling claims: guidance for industry. 2009.
- 11 5. (EMA), C.f.M.P.f.H., Reflection paper on the regulatory guidance for the use of health-related quality of life
12 (HRQL) measures in the evaluation of medicinal products. 2005.
- 13 6. Bevans K, et al. Patient reported outcomes as indicators of pediatric health care quality. *Acad Pediatrics.*
14 2014;14(5):S90-S96.
- 15 7. Brazier J, et al. Measuring and valuing health benefits for economic evaluation. 2nd ed. Oxford: University
16 Press; 2017.
- 17 8. Karimi M, Brazier J. Health, health-related quality of life, and quality of life: what is the difference?
18 *Pharmacoeconomics.* 2016; <https://doi.org/10.1007/s40273-016-0389-9>
- 19 9. Black, N. Patient reported outcome measures could help transform healthcare. *BMJ.* 2013;
20 <https://doi.org/10.1136/bmj.f167>
- 21 10. Kwon J, et al. Systematic review of conceptual, age, measurement and valuation considerations for generic
22 multidimensional childhood patient-reported outcome measures. *Pharmacoeconomics.* 2022;
23 <https://doi.org/10.1007/s40273-021-01128-0>
- 24 11. Janssens A, et al. A systematic review of generic multidimensional patient-reported outcome measures for
25 children, part I: descriptive characteristics. *Value Health.* 2015; <https://doi.org/10.1016/j.jval.2014.12.006>

- 1 12. Meryk A, et al. Implementation of daily patient-reported outcome measurements to support children with
2 cancer. *Pediatric Blood & Cancer*. 2021; <https://doi.org/10.1002/pbc.29279>
- 3 13. Arbuckle R, Abetz-Webb L. “Not just little adults”: qualitative methods to support the development of
4 pediatric patient-reported outcomes. *The Patient-Patient-Centered Outcomes Research*. 2013;
5 <https://doi.org/10.1007/s40271-013-0022-3>
- 6 14. Patrick D, et al. Content validity—establishing and reporting the evidence in newly developed patient-reported
7 outcomes (PRO) instruments for medical product evaluation: ISPOR PRO Good Research Practices Task Force
8 report: part 2—assessing respondent understanding. *Value Health*. 2011;
9 <https://doi.org/10.1016/j.jval.2011.06.013>
- 10 15. Patrick D, et al. Content validity—establishing and reporting the evidence in newly developed patient-reported
11 outcomes (PRO) instruments for medical product evaluation: ISPOR PRO good research practices task force
12 report: part 1—eliciting concepts for a new PRO instrument. *Value Health*. 2011;
13 <https://doi.org/10.1016/j.jval.2011.06.014>
- 14 16. Terwee CB, et al. COSMIN methodology for evaluating the content validity of patient-reported outcome
15 measures: a Delphi study. *Quality Life Res*. 2018; <https://doi.org/10.1007/s11136-018-1829-0>
- 16 17. Brod M, Tesler LE, Christensen TL. Qualitative research and content validity: developing best practices based
17 on science and experience. *Quality Life Res*. 2009; <https://doi.org/10.1007/s11136-009-9540-9>
- 18 18. Lasch K, et al. PRO development: rigorous qualitative research as the crucial foundation. *Quality Life Res*.
19 2010; <https://doi.org/10.1007/s11136-010-9677-6>
- 20 19. Stevens K, Palfreyman S. The use of qualitative methods in developing the descriptive systems of preference-
21 based measures of health-related quality of life for use in economic evaluation. *Value Health*. 2012;
22 <https://doi.org/10.1016/j.jval.2012.08.2204>
- 23 20. Willis GB. *Cognitive interviewing: A tool for improving questionnaire design*. Sage Publications; 2005
- 24 21. Terwee CB, et al. *COSMIN methodology for assessing the content validity of PROMs—user manual*.
25 Amsterdam: VU University Medical Center, 2018.
- 26 22. Patel ZS, Jensen SE, Lai -S. Considerations for conducting qualitative research with pediatric patients for the
27 purpose of PRO development. *Quality Life Res*, 2016; <https://doi.org/10.1007/s11136-016-1256-z>

- 1 23. Arsiwala T, et al. Measuring What Matters for Children: A Systematic Review of Frequently Used Pediatric
2 Generic PRO Instruments. *Therapeutic innovation & regulatory science*. 2021; [https://doi.org/10.1007/s43441-](https://doi.org/10.1007/s43441-021-00311-x)
3 [021-00311-x](https://doi.org/10.1007/s43441-021-00311-x)
- 4 24. Husbands S, Mitchell PM, Coast J. A systematic review of the use and quality of qualitative methods in
5 concept elicitation for measures with children and young people. *The Patient-Patient-Centered Outcomes*
6 *Research*. 2020; <https://doi.org/10.1007/s40271-020-00414-x>
- 7 25. Willis J, et al. Engaging the Voices of Children: A Scoping Review of How Children and Adolescents Are
8 Involved in the Development of Quality-of-Life-Related Measures. *Value Health*. 2021;
9 <https://doi.org/10.1016/j.jval.2020.11.007>
- 10 26. Rothmund M, et al. A critical evaluation of the content validity of patient-reported outcome measures assessing
11 health-related quality of life in children with cancer: a systematic review. *J Patient-Reported Outcomes*. 2023;
12 <https://doi.org/10.1186/s41687-023-00540-8>
- 13 27. Basic documents: forty-ninth edition (including amendments adopted up to 31 May 2019). Geneva: World
14 Health Organization; 2020. Licence: CC BY-NC-SA 3.0 IGO
- 15 28. Page MJ, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ*.
16 2021; <https://doi.org/10.1136/bmj.n71>
- 17 29. Hyslop S, et al. Identifying symptoms using the drawings of 4–7 year olds with cancer. *Eur J Oncol Nurs*.
18 2018; <https://doi.org/10.1016/j.ejon.2018.08.004>
- 19 30. Tomlinson D, et al. Development of mini-SSPedi for children 4-7 years of age receiving cancer treatments.
20 *BMC Cancer*. 2019; <https://doi.org/10.1186/s12885-018-5210-z>
- 21 31. McHugh ML. Interrater reliability: the kappa statistic. *Biochemia medica*. 2012;22(3);276-282.
- 22 32. Carlton J. Identifying potential themes for the Child Amblyopia Treatment Questionnaire. *Optom Vis Sci*.
23 2013; <https://doi.org/10.1097/OPX.0b013e318290cd7b>
- 24 33. Carlton J. Developing the draft descriptive system for the child amblyopia treatment questionnaire (CAT-
25 Qol): a mixed methods study. *Health & Quality of Life Outcomes*. 2013; [https://doi.org/10.1186/1477-7525-11-](https://doi.org/10.1186/1477-7525-11-174)
26 [174](https://doi.org/10.1186/1477-7525-11-174)

- 1 34. Connor NP, et al. Attitudes of children with dysphonia. *J Voice*. 2008;
2 <https://doi.org/10.1016/j.jvoice.2006.09.005>
- 3 35. Zieschank KL, et al. Children's Perspectives on Emotions Informing a Child-Reported Screening Instrument.
4 *J. Child Fam Stud*. 2021; <https://doi.org/10.1007/s10826-021-02086-z>
- 5 36. Coussens M. et al. A Qualitative Photo Elicitation Research Study to elicit the perception of young children
6 with Developmental Disabilities such as ADHD and/or DCD and/or ASD on their participation. *PLoS ONE*. 2020;
7 <https://doi.org/10.1371/journal.pone.0229538>
- 8 37. Christie D, et al. Exploring views on satisfaction with life in young children with chronic illness: an innovative
9 approach to the collection of self-report data from children under 11. *Clin Child Psychol Psychiatry*. 2012;
10 <https://doi.org/10.1177/1359104510392309>
- 11 38. Follansbee-Junger KW, et al. Development of the PedsQL (TM) Epilepsy Module: Focus group and cognitive
12 interviews. *Epilepsy & Behav*. 2016; <https://doi.org/10.1016/j.yebeh.2016.06.011>
- 13 39. Franciosi JP, et al. Quality of life in paediatric eosinophilic oesophagitis: what is important to patients? *Child
14 Care Health Dev*. 2012; <https://doi.org/10.1111/j.1365-2214.2011.01265.x>
- 15 40. Krenz U, et al. Health-related quality of life after pediatric traumatic brain injury: A qualitative comparison
16 between children's and parents' perspectives. *Plos One*. 2021; <https://doi.org/10.1371/journal.pone.0246514>
- 17 41. Nutakki K, et al. Development of the pediatric quality of life inventory neurofibromatosis type 1 module items
18 for children, adolescents and young adults: qualitative methods. *J Neuro-Oncol*. 2017;
19 <https://doi.org/10.1007/s11060-016-2351-2>
- 20 42. Panepinto JA, Torres S, Varni JW. Development of the PedsQL TM Sickle Cell Disease Module items:
21 qualitative methods. *Qual Life Res*. 2012; <https://doi.org/10.1007/s11136-011-9941-4>
- 22 43. Varni JW, et al. PedsQL Gastrointestinal Symptoms Module Item Development: Qualitative Methods. *J
23 Pediatr Gastroenterol Nutr*. 2012; <https://doi.org/10.1097/MPG.0b013e31823c9b88>
- 24 44. Wiener L., et al. Child and parent perspectives of the chronic graft-versus-host disease (cGVHD) symptom
25 experience: a concept elicitation study. *Support. Care Cancer*. 2014; <https://doi.org/10.1007/s00520-013-1957-6>
- 26 45. Morris C, et al. Development of the Oxford ankle foot questionnaire: finding out how children are affected by
27 foot and ankle problems. *Child: Care, Health & Dev*. 2007; <https://doi.org/10.1111/j.1365-2214.2007.00770.x>

- 1 46. Markham C, et al. Children with speech, language and communication needs their perceptions of their quality
2 of life. *Int J Lang Commun Disord.* 2009; <https://doi.org/10.1080/13682820802359892>
- 3 47. Gao W, et al. Development and Pilot Testing a Self-Reported Pediatric PROMIS App for Young Children
4 Aged 5-7 Years. *J Pediatr Nurs.* 2020; <https://doi.org/10.1016/j.pedn.2020.04.003>
- 5 48. Hwang M, et al. Development of the Pediatric Quality of Life Inventory™ Spinal Cord Injury (PedsQL™
6 SCI) module: qualitative methods. *Spinal Cord.* 2020; <https://doi.org/10.1038/s41393-020-0450-6>
- 7 49. Churruca K, et al. Patient-reported outcome measures (PROMs): A review of generic and condition-specific
8 measures and a discussion of trends and issues. *Health Expect.* 2021; <https://doi.org/10.1111/hex.13254>
- 9 50. Riley AW. Evidence that school-age children can self-report on their health. *Ambul Pediatr.* 2004;
10 <https://doi.org/10.1367/A03-178R.1>
- 11 51. Curtin C. Eliciting children's voices in qualitative research. *The Am. J Occup Therap.* 2001;
12 <https://doi.org/10.5014/ajot.55.3.295>
- 13 52. Kitto S, et al. Quality in qualitative research. *Med J Aust.* 2008.
- 14 53. Boeije, H. and Willis, G. The cognitive interviewing reporting framework (CIRF). *Methodology.* 2013;
15 <https://doi.org/10.1027/1614-2241/a000075>
- 16 54. Wright J, Moghaddam N, Dawson DL. Cognitive interviewing in patient-reported outcome measures: A
17 systematic review of methodological processes. *Qualitative Psychology.* 2021;
18 <https://doi.org/10.1037/qup0000145>
- 19 55. Bevans K, et al. Conceptual and methodological advances in child-reported outcomes measurement. *Expert*
20 *Rev Pharmacoeconomics & Outcomes Res.* 2010; <https://doi.org/10.1586/erp.10.52>
- 21 56. Rebok G, et al. Elementary school-aged children's reports of their health: a cognitive interviewing study. *Qual*
22 *Life Res.* 2001; <https://doi.org/10.1023/A:1016693417166>
- 23 57. Drennan J. Cognitive interviewing: verbal data in the design and pretesting of questionnaires. *J Adv Nurs.*
24 2003; <https://doi.org/10.1046/j.1365-2648.2003.02579.x>
- 25 58. Papaioannou D, Sutton A, and Booth A. Systematic approaches to a successful literature review. 2nd ed.
26 London: SAGE Publications; 2016

1 59. Peters M, et al. Guidance for conducting systematic scoping reviews. JBI Evid Implement. 2015;
2 <https://doi.org/10.1097/XEB.0000000000000050>

3 60. Campbell F, et al. Mapping reviews, scoping reviews, and evidence and gap maps (EGMs): the same but
4 different – the “Big Picture” review family. Systematic Rev. 2023; <https://doi.org/10.1186/s13643-023-02178-5>

5

6

7

8

9

10

11

12

13

14

15

16

17

18

19

20

21

22

23

1 **Footnotes**

2 1. A scoping review aims to systematically identify the breadth of evidence on a particular topic and to describe
3 the characteristics of that evidence [58, 60]. Given this area of research is relatively unknown, especially within
4 the specific age range of ≤ 7 years, a scoping review methodology was considered appropriate.

5

1 *Tables*

2 Table 1. Example search strategy used for Medline via Ovid. All search terms were free text unless otherwise
3 stated.

| | Concept | Search terms |
|---|--|--|
| 1 | Population (children ≤7 years) | Child (<i>MESH</i>) OR child health (<i>MESH</i>) OR “children” |
| 2 | PRO instruments (standardised questionnaires that measure subjective health from a patient’s own perspective without interpretation by others) | “patient reported outcome*” OR “instrument” OR “measure” |
| 3 | Development and/or testing of PROMs, or development of a conceptual framework for a subjective health concept | “develop*” OR “content validity” OR “concept elicitation” OR “cognitive” |
| 4 | Qualitative methods – methods that collect qualitative data and used qualitative analysis techniques | Qualitative research (<i>MESH</i>) OR “interview*” OR “focus group*” |
| | | 1 AND 2 AND 3 AND 4 |

4 Abbreviations: MESH (Medical Subject Heading)

5

6

7 Table 2. Methodological recommendations identified from guidance documents [3, 13, 22].

| Type of recommendation | Recommendation |
|--|---|
| Topic/method guide | <ul style="list-style-type: none"> Explain procedure to children Consider question wording e.g., avoid double negatives Consider presentation of questions e.g., direct/open first Consider question content e.g., focus on concrete concepts Pilot test method Adapt topic guide to meet needs across sessions |
| Practical considerations during data collection | <ul style="list-style-type: none"> Consider role of parents Interviewer communication e.g., mitigate social desirability, reduce power imbalance Build rapport before session Consider setting as age appropriate Consider length of session as age appropriate Consider group size regarding age Consider group dynamics regarding age Show and explain recording equipment Assess child’s level of understanding of the session Ensure facilitators are trained |
| Additional/modified activities | <ul style="list-style-type: none"> Use enabling activities e.g., drawing, photo elicitation, participatory activities Warm-up activities Option for alternative or creative ways in which children can respond to questions |
| Cognitive interviewing recommendations | <ul style="list-style-type: none"> Follow same general procedure as with adults Conduct cognitive testing within narrow age bands Give children time to respond Pay close attention to a child’s understanding of recall period Consider age and length of interview Explain interview process and child’s role Consider content of questions e.g., not too abstract or hypothetical |

8

1 Table 3. Aspects of PROMs and aspects of content validity recommended to be assessed in cognitive interviews
 2 by ISPOR [14] and COSMIN [21]
 3

| Aspect of PROM/content validity recommended to be assessed | ISPOR recommendation | COSMIN recommendation |
|--|----------------------|-----------------------|
| Comprehensibility of instructions (how do respondents interpret the task?) | ✓ | ✓ |
| Comprehensibility of items (are the items clear for respondents? How do they interpret the meaning of items?) | ✓ | ✓ |
| Comprehensibility of recall period (how do respondents retrieve and remember information?) | ✓ | ✓ |
| Comprehensibility of response options (are the different response levels clear for respondents? How do respondents interpret the different options?) | ✓ | ✓ |
| Comprehensiveness (does the PROM contain all relevant concepts for the target population?) | ✓ | ✓ |
| Format (is the presentation of the PROM clear for respondents?) | ✓ | |
| Length (does the time taken to complete the PROM place burden on respondents?) | ✓ | |

4

5

1 Table 4. Study characteristics

| Author, year, reference | Title | Aim/s of the paper | Specific condition or domain | Age range (years) | Sample size ≤7 years | Element of PROM development | Subjective health concept | Country of research |
|-------------------------------------|---|---|---|-------------------|----------------------|--|---------------------------|---------------------|
| Carlton, 2013 [32] | Identifying potential themes for the child amblyopia treatment questionnaire | This was one of several papers reporting the developing of a PROM for amblyopia for children aged 4-7 years (CAT-QoL). This paper reported the identification of potential themes and items for the CAT-QoL from conducting semi-structured interviews with children. | Amblyopia treatment | 3-9 | 54 | Concept elicitation | HRQoL/QoL | UK |
| Carlton, 2013 [33] | Developing the draft descriptive system for the child amblyopia treatment questionnaire (CAT-QoL): a mixed methods study | This was one of several papers reporting the developing of a PROM for amblyopia for children aged 4-7 years (CAT-QoL). This paper reported the use of qualitative and quantitative methods to refine the descriptive system of the CAT-QoL. | Amblyopia treatment | 5-9 | 27 | Cognitive testing | HRQoL/QoL | UK |
| Christie et al, 2011 [37] | Exploring views on satisfaction with life in young children with chronic illness: An innovative approach to the collection of self-report data from children under 11 | To describe the development of a pilot data collection tool that will inform the future development of a self-report measure of life satisfaction for children aged 3-11 years | Chronic illness e.g., diabetes, juvenile idiopathic arthritis | 3-11 | Unclear | Concept elicitation | HRQoL/QoL | UK |
| Connor et al, 2011 [34] | Attitudes of children with dysphonia | To determine the impact of voice disorders on the lives of children from the perspective of children and their caregivers | Dysphonia | 2-18 | 20 | Concept elicitation | HRQoL/QoL | USA |
| Coussens et al, 2020 [36] | A qualitative photo elicitation research study to elicit the perception of young children with developmental disabilities such as ADHD and/or DCD and/or ASD on their participation | To investigate how young children with developmental disabilities define their participation, recognising their right to express their preferences and feelings, and be consulted on matters that affect them | Developmental disabilities e.g., ADHD | 5-9 | 15 | Concept elicitation | Participation | Belgium |
| Follansbee-Junger et al, 2016 [38]† | Development of the PedsQL epilepsy module: focus group and cognitive interviews | To create an epilepsy-specific module of the PedsQL that will build on existing measures. This paper reports the first 3 steps in the validation process - how the items were generated, modified, and adapted based on a thorough literature review, expert feedback, focus groups and cognitive interviewing with children with epilepsy and their caregivers | Epilepsy | 5-18 | Unclear | Concept elicitation (and content generation) | HRQoL/QoL | USA |

| | | | | | | | | |
|----------------------------|---|--|---|------|---------|---------------------|---------------------------|---------|
| Franciosi et al, 2012 [39] | Quality of life in paediatric eosinophilic oesophagitis: what is important to patients? | To conduct focus interviews of paediatric patients with eosinophilic oesophagitis (EoE) and their parents to identify the key EoE disease specific HRQoL concerns | Eosinophilic oesophagitis | 5-18 | Unclear | Concept elicitation | HRQoL/QoL | USA |
| Gao et al, 2020 [47] | Development and pilot testing a self-reported paediatric PROMIS app for young children aged 5-7 years | To use PROMIS methods to develop a smartphone app to monitor emotional distress of children aged 5-7 years. This paper reports the testing of the usability of the app and exploration of the level of agreement between child and parent reports | Domain-specific – psychological functioning among a group of hospitalised children e.g., cancer and pneumonia | 5-7 | 15 | Cognitive testing | Psychological functioning | China |
| Hwang et al, 2020 [48] | Development of the paediatric quality of life inventory Spinal cord injury (PedsQL SCI) module: qualitative methods | Overall aim is to develop a disease-specific module of the PedsQL for SCI. This paper reports the methods and results of the qualitative research | Spinal cord injury | 5-25 | 3 | Cognitive testing | HRQoL/QoL | USA |
| Hyslop et al, 2018 [29] | Identifying symptoms using the drawings of 4–7-year-olds with cancer | To describe how children with cancer aged 4-7 years express their symptoms through drawing (a sub-component of a larger study that aimed to develop a symptom screening tool, the mini-SSPedi) | Cancer | 4-7 | 30 | Concept elicitation | Symptoms | Canada |
| Krenz et al, 2021 [40] | Health-related quality of life after paediatric traumatic brain injury: a qualitative comparison between children’s and parent’s perspectives | To provide original material from small group interviews with individuals after traumatic brain injury (TBI) concerning what they state is relevant for their HRQoL. To initially explore similarities and differences between children’s and parent’s reports. Part of a larger study aiming to adapt a TBI-specific HRQoL questionnaire (QOLIBRI) for 3 age groups (Kiddy 6-7, Kid 8-12, and Ado 13-17 years) | Traumatic brain injury | 5-17 | 4 | Concept elicitation | HRQoL/QoL | Germany |
| Markham et al, 2009 [46] | Children with speech, language, and communication needs: their perceptions of their quality of life | To provide a qualitative, child-centred, description of the quality of life experiences of children and young people with speech, language, and communication needs (SLCN). | Speech, language, and communication difficulties | 6-18 | 4 | Concept elicitation | HRQoL/QoL | UK |
| Morris et al, 2007 [45] | Development of the Oxford ankle foot questionnaire: finding out how children are affected by foot and ankle problems | To use child-centred focus group methods to identify how children’s lives are affected by foot and ankle problems | Foot and ankle problems | 5-15 | 5 | Concept elicitation | HRQoL/QoL | UK |

| | | | | | | | | |
|-----------------------------|--|---|---|------------|---------|--|---------------------------|-----------|
| Nutakki et al, 2017 [41]† | Development of the paediatric quality of life inventory neurofibromatosis type 1 module items for children, adolescents, and young adults: qualitative methods | To develop the items and support the content validity for the PedsQL NF1 module for children, adolescents, and young adults. This paper described the item development and content validation phase | Neurofibromatosis type 1 | 5-25 years | 5 | Concept elicitation (and content generation) | HRQoL/QoL | USA |
| Panepinto et al, 2012 [42]† | Development of the PedsQL sickle cell disease module items: qualitative methods | To develop the items and support the content validity of the PedsQL sickle cell disease module for paediatric patients with sickle cell disease. | Sickle cell disease | 5-18 | Unclear | Concept elicitation (and content generation) | HRQoL/QoL | USA |
| Tomlinson et al, 2019 [30] | Development of mini-SSPedi for children 4-7 years of age receiving cancer treatments | To develop a new self-report symptom screening tool for children receiving cancer treatments who are 4-7 years of aged (mini-SSPedi) based on SS-Pedi | Cancer | 4-7 | 100 | Cognitive testing | Symptoms | Canada |
| Varni et al, 2012 [43]† | PedsQL Gastrointestinal symptoms module item development: qualitative methods | To describe the qualitative methods used in the item development and content validation phase for the new PedsQL Gastrointestinal symptoms module items | Gastrointestinal disorders e.g., chronic constipation | 5-18 | 2 | Concept elicitation (and content generation) | HRQoL/QoL | USA |
| Wiener et al, 2014 [44] | Child and parent perspective of the chronic graft-versus-host disease (cGVHD) symptom experience: a concept elicitation study | To elicit description of the cGVHD symptom experience directly from children and compare the language they used to describe their symptoms and the comprehension of symptom concepts across the developmental spectrum | Chronic graft-versus host disease | 5-17 | 8 | Concept elicitation | HRQoL/QoL | USA |
| Zieschank et al, 2021 [35] | Children's perspective on emotions informing a child-reported screening instrument | To produce child-informed typologies of emotional and behavioural constructs to inform the development of a screening tool. Part of a larger study aiming to develop a self-reported digitally animated screening tool for child emotional and behavioural distress | Domain-specific – psychological functioning | 5-11 | 7 | Concept elicitation | Psychological functioning | Australia |

1

2 †These studies implied that children ≤ 7 years had also been involved in cognitive testing. However, this was not explicit and there was not enough detail of their involvement for this to be included in the analysis. They
3 are therefore classified as concept elicitation studies in this review.

4 Abbreviations: ADHD (Attention Deficit Hyperactivity Disorder); ASD (Autism Spectrum Disorder); CAT-QoL (Child Amblyopia Treatment Questionnaire); cGVHD (Chronic Graft-versus Host Disease); DCD
5 (Developmental Coordination Disorder); EoE (Eosinophilic Oesophagitis); HRQoL/QoL (Health-related quality of life/Quality of life); mini-SSPedi (Mini Symptom Screening in Pediatrics Tool); NF1
6 (Neurofibromatosis type 1); PedsQL (Pediatric Quality of Life Inventory); PROMIS (Patient Reported Outcomes Measurement Information System); QOLIBRI (Quality of Life after Brain Injury); SCI (Spinal cord
7 Injury); SLCN (Speech, language, and communication needs); TBI (Traumatic Brain Injury).

8

1 Table 5. Methods reported by concept elicitation studies

| Paper | High-level method | Authors report success?† | Enabling activity | Topic/method guide considerations (e.g., development of questions, wording of questions) | Practical considerations (e.g., setting, parent role, rapport building) | Outcomes of children's involvement (e.g., meaningfulness and use of data, author reflections) |
|--------------------------|-----------------------------------|---------------------------------|--|--|--|--|
| Carlton (2013) [32] | Individual interviews | Mixed | Drawing was used as a rapport-building activity but stopped after several interviews because it was distracting. No other enabling activities are reported. | Simple questions about child's favourite toy asked first to build rapport. Inductive approach. Interviews intended to be as open-ended as possible, children encouraged to talk about amblyopia and probes were used for further information if needed. Themes for discussion pre-determined. Verbal and non-verbal responses considered for analysis. | Individual interviews instead of focus groups because of the potential sensitive content that could arise. Parents present but not involved. Author reflects that the setting (eye clinic) may influence children's responses. Children encouraged to verbalise feelings because audio equipment would not record non-verbal cues. | Overall, author concluded that children aged 4-7 years could understand and articulate the impact of amblyopia and its treatment on their daily lives. Five interviews terminated because children were unresponsive. |
| Christie (2011) [37] | Individual interviews (3-4 years) | × | Originally a board game was developed where children answered pre-set questions about QoL. Part way through data collection, the questions were delivered to 3-4-year-olds via a storybook because it was noted that children 3-4 years struggled to answer the questions. | Reported that wording was age appropriate. Questions in the story book were the same as had been used in the board game originally – these were developed from a review of QoL measures, expert opinions, and focus groups with adolescent meningitis survivors. | Individual interviews replaced group interviews for 3-4-year-olds following pilot testing. The rationale for this is not reported. Reports that wherever possible, parents were not present. | Authors report that the story book did not increase accessibility of material for 3-4-year-olds. It is unclear whether data gathered from 3-4-year-olds is used in analysis. |
| Connor et al (2011) [34] | Individual interviews | Mixed | None reported. | Children interviewed in aged bands (2-4 years and 5-7 years) following an interview script with pre-set questions based on a preliminary conceptual framework. Follow-up probes used where needed. | Children interviewed with parents to reduce any anxiety among children or parents. A constant set of instructions was read to all children explaining that they would have a conversation about their scratchy voice. Reports rapport was built throughout the interviews. | Authors report that 2-4-year-olds gave limited information, so parent responses were primarily used for this age group. Children 5-7 years reported to provide enough information for analysis. Authors reflect that young children can talk about their awareness of and concerns about a health condition. |

| | | | | | | |
|-------------------------------------|-----------------------|--------------|--|---|---|---|
| Coussens et al (2020) [36] | Individual interviews | ✓ | <p>Photo elicitation activity – children given task of taking photos of activities at home, school, and in the community. They chose which photos to discuss in an open-ended interview.</p> <p>Informed assent procedure adapted via a comic book information sheet.</p> <p>Parents present in the interview to support children if needed and provide greater depth to the discussion.</p> | <p>Children could direct the topic of conversation by choosing the pictures they wanted to talk about. Asked to describe the pictures and prompted where needed. Used props to explain the task, child-friendly language, encouraged the child to ask questions, and explained that the child was the expert. Questions avoided ‘why’, were no more than 5 words longer than children’s utterances, and focussed on things that are observable/the immediate situation.</p> | <p>Parents were present to support the child when they had difficulties expressing their thoughts as the authors believed this would enable greater depth to be gathered from the interviews. Photo elicitation chosen because previous research suggested it was effective for involving children with communication impairments in research. Instructions to interviewers to reduce power imbalances. Played a favourite game before the interview to build rapport. Took place in the child’s home to provide a familiar and safe environment.</p> | <p>Authors concluded that it is possible for young children to accurately inform about their participation experiences; authors reflect that this is because of the method used and the adapted approach to gathering informed consent.</p> |
| Follansbee-Junger et al (2016) [38] | Individual interviews | Not reported | None reported. | All PedsQL module development studies that reported the same procedure as is typical for PedsQL studies – interviews conducted following a semi-structured interview guide that had been developed based on themes from literature review and expert opinion. | Parents present for children aged 5-7 years, but their role in the interview is not explicitly reported. | Overall limited reporting of the outcomes of interviews conducted with children 5-7 years |
| Nutakki et al (2017) [41] | | Not reported | | | | |
| Panepinto et al (2012) [42] | | Not reported | | | | |
| Varni et al (2012) [43] | | Not reported | | | | |
| Franciosi et al (2012) [39] | | ✓ | | | | |
| Hylsop et al (2018) [29] | Individual interviews | Mixed | <p>Drawing – children were given the option to draw a ‘not good’ or ‘bad’ day at the start of the interview. If they chose to draw, pictures were then discussed in the interview. Drawing chosen to alleviate anxiety,</p> | <p>Only one open-ended question was asked about the drawings – “tell me about your drawing”. This was an exploratory study to see whether drawing could support children in talking</p> | <p>Authors report only asking children to draw about one topic and to do one drawing to reduce cognitive burden. Second researcher present to record what the child said and</p> | <p>Overall authors report drawing as helping to build rapport, being engaging for children, and providing an insight into how young children conceptualise their</p> |

| | | | | | | |
|--------------------------|--------------|-------|--|---|--|--|
| | | | facilitate communication, and add further dimension to interview responses. | about their symptoms – there was no preliminary conceptual framework guiding interview questions. Avoided over probing to ensure children did not believe they had given a wrong answer. | allow the interviewer to maintain eye contact and rapport with the child. A hand puppet was also used at the start of the study to build rapport. | health – it was possible to identify four themes from the drawings. Authors reflect that a combination of methods is necessary as 12 of the 30 children did not want to draw. |
| Krenz et al (2021) [40] | Focus groups | ✓ | None reported. | Children aged 5-7 years were interviewed in a focus group about HRQoL after traumatic brain injury. Existing conceptual model of HRQoL was used to generate questions and support analysis. | Parents took part in their own interview groups. Data compared across parent and child groups to identify agreement/disagreement. | Authors conclude that children as young as 5 years could discuss their HRQoL. |
| Markam et al (2009) [46] | Focus groups | ✓ | Picture-card fishing game – pre-set picture cards depicting aspects of QoL were used in a fishing game for children aged 6-7 years to support their engagement in the discussion. Reported using this activity because similar activities are often used by speech and language therapists when working with children with communication difficulties. | Pre-set aspects of QoL for discussion were developed from a literature review. Aimed to use minimal prompts beyond this for an open-ended discussion. | Focus groups took place at the children’s school to try and maximise their comfort and confidence. Timing reported to follow guidelines in the literature of 30-45 minutes for children 6-7 years. Recruited into focus groups representing school peer groups to facilitate communication and group dynamics. | Authors reflect that the picture-cards may have increased adult-projection of QoL onto the results, but an activity like this was necessary to engage children with SLCN. Authors conclude that while quotes from 6-7-year-olds were shorter, their content was consistent with data from older children. Report that the enabling method was successful at engaging children with SLCN. |
| Morris et al (2007) [45] | Focus groups | Mixed | Statement agreement – 5-7-year-olds asked to state their agreement to statements (e.g., “having a foot problem affects what activities children can do) in any way they liked e.g., moving round the room to stand next to statements they agreed with. Life-mapping activity – 5-7-year-olds asked to talk about the effects a foot and ankle problem might have on a child at different times of the day. | Statements started broad then became more specific as the session progressed. Life-mapping activity reported to have been chosen to depersonalise the issues that were discussed. | Children could choose how they wanted their life map recorded – 5-7-year-olds chose to use a large sheet of wallpaper with the facilitator transcribing their thoughts. Parents took part in separate focus groups. Emphasised that there was no right or wrong answer and that some people would have different answers, warm-up activity at the start of the session and all made name badges before, had an | Authors report that the enabling activities were successful at eliciting children’s experiences of having a foot or ankle problem. Authors reflect that the warm-up helped put children at ease and that the facilitator did a greater proportion of children in younger age groups. Authors concluded that children’s ability to participate depends on individual maturity as |

| | | | | | | |
|-----------------------------|---------------------------------------|--------------|---|--|--|---|
| | | | | | additional receptionist to greet the participants and wait outside in case any children became distressed, snacks and drinks provided, moved the microphone to young children could sit on the floor and move around the room. | opposed to chronological age. |
| Wiener et al (2014) [44] | Individual interviews | Not reported | Parents interviewed with children aged 5-7 years to support greater depth in the discussion and to mitigate the potential of recency effects | Interview questions were pre-defined based on a literature review and the symptom experience of adults. Open-ended questions were used, and children were encouraged to identify concerns in specific content areas e.g., skin | Children 5-7 years interviewed with parents because they were anticipated to have more limited vocabulary and understanding of health concepts. | Authors reflect that prompts were needed for children 5-7 years to report when a recall period extending beyond the past day was used. |
| Zieschank et al (2021) [35] | Focus groups or individual interviews | ✓ | Multi-sensory response options were used. Children 5-6 years were asked about emotion constructs – they gave a verbal description, provided a facial expression, audio cue, and behavioural cue of the emotion, and described a story of that emotion. Children could choose to be in a group or to be in an individual interview. | Emotion constructs discussed were pre-defined, identified from existing instruments for adults and refined through a Delphi panel of nine child researchers and psychologists. Facilitator used minimal prompts to stimulate discussion and encourage the sharing of narratives. | Posters of the emotions were included around the room to allow children who did not want to talk to point at an idea. Rapport was built before the focus group started and children could choose if they wanted their parent present. Facilitator sat in a circle with the children using child-sized chairs. Focus groups composed of friends or siblings to maximise participation and ensure comfort. | Authors reflected that obtaining data from multiple categories (i.e., verbal description, behavioural cues, narratives etc.) gave them a lot of information to draw from during concept elicitation. Reflect that the child-participatory methods helped give children a voice. Also reflected that younger children were less verbally expressive. |

1 Abbreviations: HRQoL (Health-related quality of life); PedsQL (Pediatric Quality of Life Inventory); QoL (Quality of Life)

2 †Success defined by authors of included studies as children participating in the qualitative activities and/or providing meaningful data

1 Table 7. Methods reported by cognitive interviewing studies

| Paper | High-level method | Authors report success?† | Enabling activity | Topic/method guide considerations (e.g., development of questions, wording of questions) | Practical considerations (e.g., setting, parent role, rapport building) | Outcomes of children's involvement (e.g., meaningfulness and use of data, author reflections) |
|-----------------------------|--------------------------|---------------------------------|--|--|--|--|
| Carlton (2013) [33] | Individual interviews | ✓ | None reported. | Cognitive interviews conducted whereby children read and completed the questionnaire, were asked to explain what they thought the questions were asking them and were asked their opinion of the layout. Observations were made of the child's ability to read the questionnaire. | If it was apparent that the child did not understand the task, the interview was ended. | Overall author concluded that research with young children could support the face and content validity of an instrument and that the data gathered was useful in refining the instrument. |
| Gao et al (2020) [47] | Individual interviews | Not reported | None reported. | Children completed the questionnaire and were asked standardised questions about the clarity of instructions, items, recall period, and response options. They were asked to explain why they chose certain answers and to describe what they thought about while completing the questionnaire. Asked if any items were not relevant or offensive. | None reported. | Authors do not reflect explicitly on the data provided by children. |
| Hwang et al (2020) [48] | Individual interviews | Not reported | None reported. | Instrument was administered via an interviewer and verbal probes were used after each item to assess the child's level of understanding. | Reports that interviews were conducted separately for children and for parents. | Results were used to modify the wording of items for the instrument for 5-7-year-olds. Domains were removed that children did not understand. |
| Tomlinson et al (2019) [30] | Individual interviews | ✓ | Multi-stage approach to cognitive interviewing used to limit cognitive burden on children 4-7 years. Clarity of recall period, response options, and understanding of the word 'bother' were assessed first. When modified and at least 80% of children understood these concepts, the clarity of individual items was assessed. When modified and at least 80% of children understood, the entire PROM was presented to children. | When assessing response options, began by testing understanding of a dichotomous response, then 3-point scales. Children also asked for their preference of pictorial response options. When assessing clarity of individual symptoms, these were presented one at a time in a windowed frame board and verbal probes were used. When presented with the entire PROM, children were asked to judge ease of completion through a 3-point Likert scale ("too short", "just right", or "too long"). Started with direct questions that were easy to answer then progressed to open-ended questions. | Children could choose to have the questionnaire read out loud to them. Determined children's understanding of 'yesterday/today' by asking them what they did and asked parents to confirm this. Multi-stage approach had been used successfully before with older children. Clarified the purpose of the interview at the start. Non-verbal actions were also recorded. Parents were present but asked not to intervene in the interview. A puppet was used to engage the child. | Data from cognitive interviews was used to modify the PROM e.g., the incorporation of synonyms. Authors report that a more phased approach to cognitive interviewing is needed with younger children. Most children chose to have the questionnaire read aloud to them. Authors report that a large sample was needed to produce a satisfactory version of the PROM. |

†Success defined by authors of included studies as children participating in the qualitative activities and/or providing meaningful data

- 1 Table 8. Aspects of content validity and PROM elements (identified from ISPOR content validity guidance to support the conduct of cognitive interviews [14] and COSMIN
- 2 risk of bias checklists for the evaluation of PROM development and content validity studies [21]) assessed in cognitive testing studies

| Authors, reference, and year | Title | Subjective health concept | Aspects of content validity and PROMs assessed in cognitive interviews | | | | | | | |
|------------------------------|--|--|--|-------------------------|--------------------------|-----------------------------|-------------------|-----------|--------|--------|
| | | | Clarity of items | Clarity of instructions | Clarity of recall period | Clarity of response options | Comprehensiveness | Relevance | Format | Length |
| Carlton, 2013 [33] | Developing the draft descriptive system for the child amblyopia treatment questionnaire (CAT-QoL): a mixed methods study | HRQoL/QoL related to amblyopia treatment | ✓ | | | | | ✓ | | ✓ |
| Gao et al, 2020 [47] | Development and pilot testing a self-reported pediatric PROMIS app for young children aged 5-7 years | Psychological functioning | ✓ | ✓ | ✓ | ✓ | | ✓ | | |
| Hwang et al, 2020 [48] | Development of the pediatric quality of life inventory Spinal cord injury (PedsQL SCI) module: qualitative methods | HRQoL/QoL related to SCI | ✓ | | | | | ✓ | | |
| Tomlinson et al, 2019 [30] | Development of mini-SSPedi for children 4-7 years of age receiving cancer treatments | Symptoms with cancer | ✓ | | ✓ | ✓ | | | | ✓ |

3 Abbreviations: SCI (Spinal cord Injury).

1 *Figure Legends*

2 **Fig. 1** Typical PROM development process informed by FDA guidance [4] and Arbuckle & Abetz-Webb [13]

3 **Fig. 2** PRISMA flow diagram [29] for database searches, reference list searches, and forward citation searches for
4 qualitative studies that involved children ≤ 7 years in PROM development/testing or concept elicitation research

5 **Fig. 3** Bar chart showing the youngest ages from which children were involved in studies

6 **Fig. 4** Frequencies of studies exploring each subjective health concept, differentiated by study type: concept
7 elicitation (CE) and cognitive interviewing (CI)

8

9

10

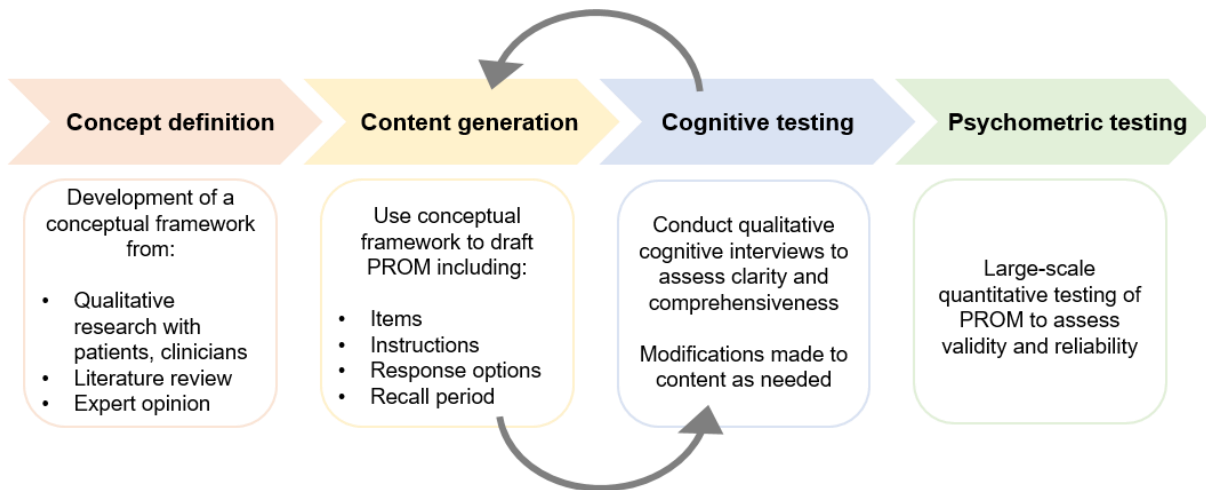
11

12

13

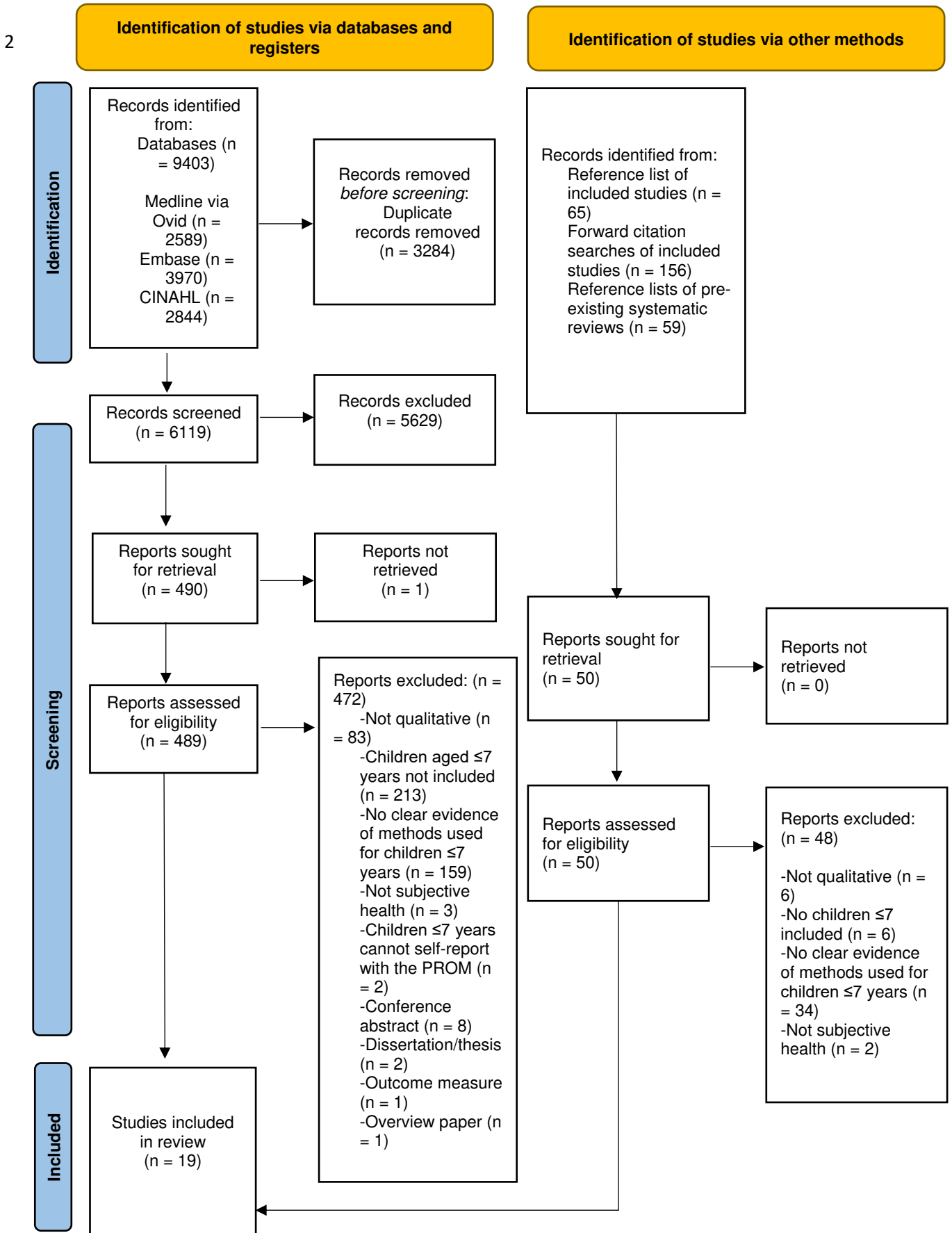
14

15 *Figure 1*

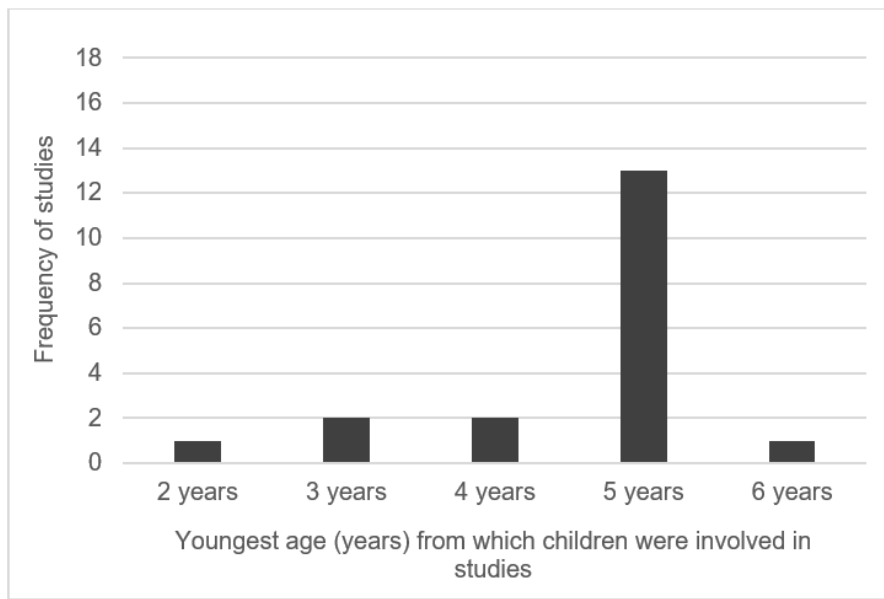


16

1 Figure 2



1 *Figure 3*



2

1 *Figure 4*

