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

Running heading: Scoping review of young children's involvement in qualitative research for PROM development

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Abstract

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

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

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

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

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

Key Points for Decision Makers

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

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

1 Introduction

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

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

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

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

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

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

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

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

The known challenges of involving young children in qualitative research, limited guidance, few examples in the literature, and broad focus of previous reviews on childhood (i.e., 0-18 years) means it is unclear whether it is possible to enable children ≤7 years to participate meaningfully in qualitative research for PROM development/testing. This scoping review aims to investigate the involvement of children aged ≤7 years in qualitative research for PROM development/testing[1]. It asks the following questions:

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

2 Methods

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

2.1 Search strategy

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

conceptual framework for a subjective health concept without explicitly stating intentions to develop a PROM. These studies were included given development of a conceptual framework is a key stage in PROM development (see Introduction and Figure 1). The search was not limited to any specific health condition and could include physical and/or mental conditions. For this review 'subjective health' was defined as any self-report coming directly from the child of their experiences of health conditions and/or treatment (e.g., symptoms, pain) or their perceptions of the impact of the health/treatment on their daily life (e.g., cognitive, emotional, or social implications). This could include physical, mental, and/or social domains, in-line with the World Health Organisation definition of health [27]. It did not include, for example, children's reports of experiences in hospital.

Medline via Ovid was used to develop search terms combining four key concepts (Table 1). The search strategy was then adapted for Embase and CINAHL (see Online Resource 2 for full search strategies for each database). Databases were searched without date limits. Forward citation searches were conducted using the "find citing articles" feature of Medline and PubMed. Reference lists of included studies and of four pre-existing systematic reviews [10, 23-25] were searched by hand.

2.2 Identifying studies for inclusion

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

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

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

2.3 Data extraction

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

2.4 Synthesis methods

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

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

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

3 Results

3.1 Search results

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

3.2 Study characteristics

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

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

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

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

3.3 Subjective health concepts

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

3.4 Qualitative methods and comparison to methodological guidance

3.4.1 Concept elicitation research

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

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

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

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

3.4.2 Cognitive interviews

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

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

4 Discussion

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

4.1 Concept elicitation studies

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

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

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

4.2 Cognitive interviewing studies

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

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

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

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

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

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

Conclusion

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

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

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Footnotes

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

Tables

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

	Concept	Search terms
1	Population (children ≤7 years)	Child (*MESH*) OR child health (*MESH*) 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 (*MESH*) OR "interview*" OR "focus group*"
		1 AND 2 AND 3 AND 4

Abbreviations: MESH (Medical Subject Heading)

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

Type of recommendation	Recommendation
Topic/method guide	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	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	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	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

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

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?)	✓	

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

†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 are therefore classified as concept elicitation studies in this review.

Abbreviations: ADHD (Attention Deficit Hyperactivity Disorder); ASD (Autism Spectrum Disorder); CAT-QoL (Child Amblyopia Treatment Questionnaire); cGVHD (Chronic Graft-versus Host Disease); DCD (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 (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 Injury); SLCN (Speech, language, and communication needs); TBI (Traumatic Brain Injury).

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	✓	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. Informed assent procedure adapted via a comic book information sheet. Parents present in the interview to support children if needed and provide greater depth to the discussion.	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.	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.	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.
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]		✓			Parents present for children aged 5-7 years but not involved in interview itself. After their child's interview they were asked for their opinion on what their child had said.	Authors reflect that it was important to speak to the children themselves and not just their parents because they often had different concerns and reflected different aspects of HRQoL.
Hylsop et al (2018) [29]	Individual interviews	Mixed	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,	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	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	Overall authors report drawing as helping to build rapport, being engaging for children, and providing an insight into how young children conceptualise their

			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 6. Methodological recommendations identified from guidance documents [3, 13, 22] reported by included studies.

Authors, reference, and year	Topic/method guide recommendations						Practical considerations during data collection									Additional or modified activities			Cognitive interviewing recommendations						
	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*	*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*	*Use enabling activities e.g., drawing, photo elicitation*	*Warm-up activities*	*Option for alternative or creative ways in which children can respond to questions*	*Follow same general procedure as with adults*	*Conduct cognitive testing within narrow age bands*	*Give children time to respond*	*Pay close attention to children'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*
Concept elicitation studies																									
Carlton, 2013 [32]	✓		✓	✓			✓		✓			N/A	✓			✓	✓	✓	N/A						
Christie et al, 2011 [37]		✓	✓	✓	✓	✓	✓					N/A				✓									
Connor et al, 2011 [34]	✓						✓		✓			N/A				✓									
Coussens et al, 2020 [36]	✓	✓	✓	✓			✓	✓	✓	✓		N/A			✓	✓	✓	✓	✓	✓					
Follansbee-Junger et al, 2016 [38]							✓																		
Franciosi et al (2012) [39]							✓	✓								✓									

Hyslop et al, 2018 [29]	✓		✓	✓	N/A *			✓	✓		✓	N/A			✓		✓	✓											
Krenz et al, 2021 [40]							✓							✓		✓													
Markham et al, 2009 [46]			✓	✓		✓		✓		✓	✓		✓			✓	✓												
Morris et al, 2007 [45]			✓				✓	✓	✓	✓			✓			✓	✓	✓	✓										
Nutakki et al, 2017 [41]							✓					N/A				✓													
Panepinto et al, 2012 [42]			✓				✓					N/A				✓													
Varni et al, 2012 [43]			✓				✓					N/A				✓													
Wiener et al, 2014 [44]							✓					N/A																	
Zieschank et al, 2021 [35]	✓						✓	✓	✓	✓			✓			✓	✓	✓	✓										
Cognitive interviewing studies																													
Carlton, 2013 [33]												N/A								✓	✓		**						
Gao et al, 2020 [47]												N/A								✓	✓								
Hwang et al, 2020 [48]			✓				✓					N/A								✓	✓								
Tomlinson et al, 2019 [30]	✓	✓	✓				✓					N/A			✓	✓	✓			✓	✓		✓	✓	✓				

*N/A as this study was exploratory. **Did not formally assess children's understanding of the recall period but a justification for the recall period is given based on children's more limited understanding of time and nature of the amblyopia treatment being assessed.

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

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 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	✓		✓	✓				✓

Abbreviations: SCI (Spinal cord Injury).

Figure Legends

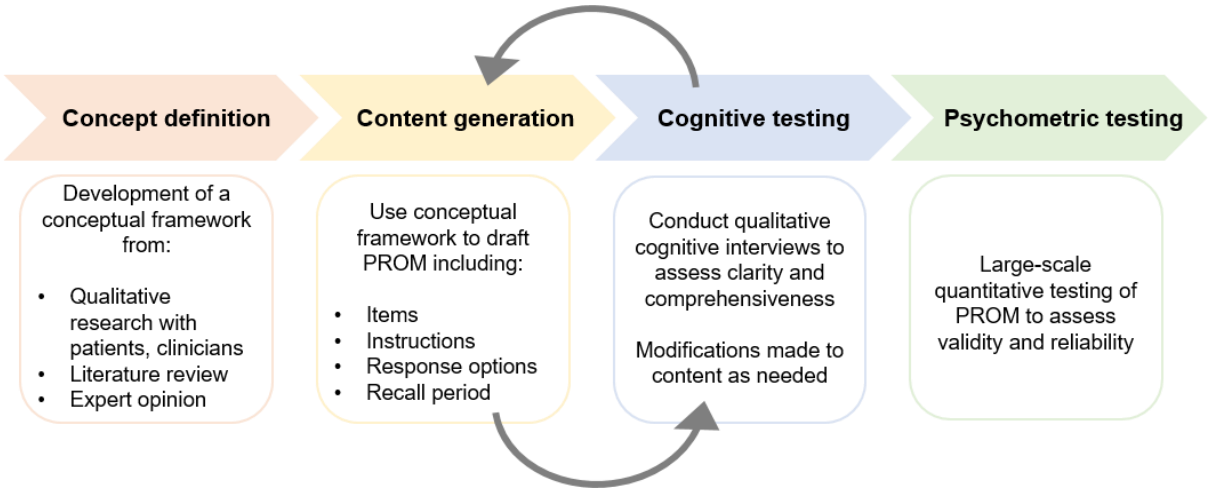
Fig. 1 Typical PROM development process informed by FDA guidance [4] and Arbuckle & Abetz-Webb [13]

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

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

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

Figure 1



1 *Figure 2*

2

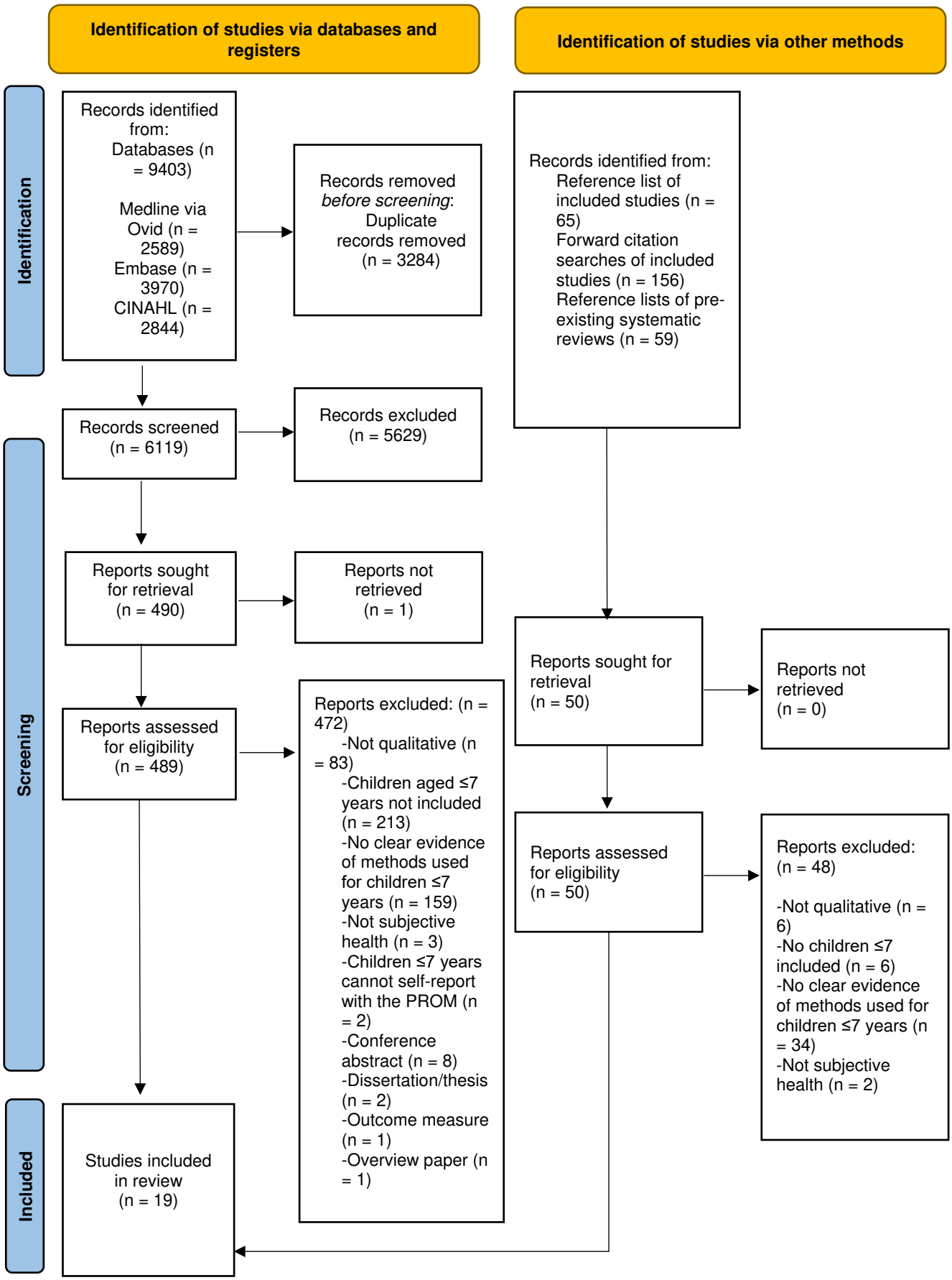
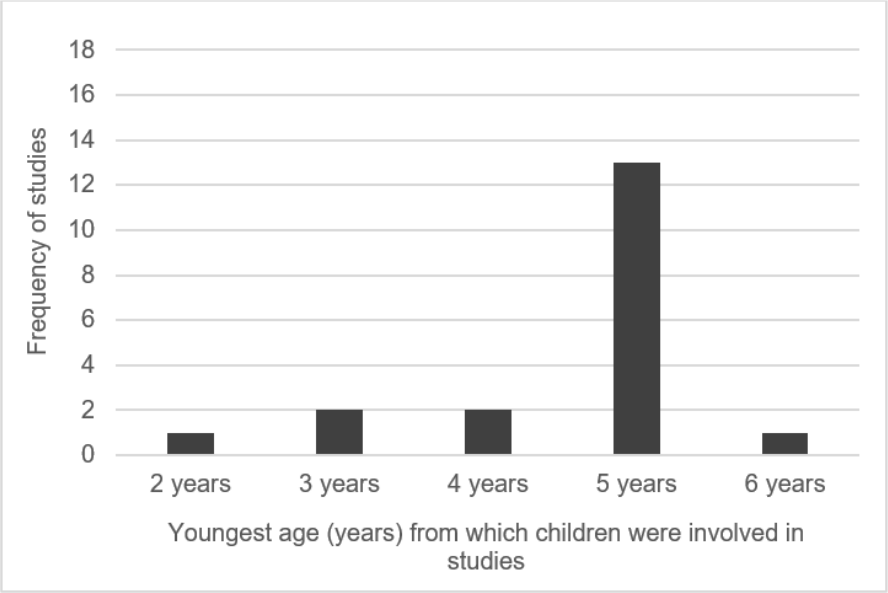

Identification of studies via databases and registers
Identification of studies via other methods
Identification
Screening
Included
Records identified from: Databases (n = 9403) Medline via Ovid (n = 2589) Embase (n = 3970) CINAHL (n = 2844)
Records removed *before screening*: Duplicate records removed (n = 3284)
Records identified from: Reference list of included studies (n = 65) Forward citation searches of included studies (n = 156) Reference lists of pre-existing systematic reviews (n = 59)
Records screened (n = 6119)
Records excluded (n = 5629)
Reports sought for retrieval (n = 490)
Reports not retrieved (n = 1)
Reports sought for retrieval (n = 50)
Reports not retrieved (n = 0)
Reports assessed for eligibility (n = 489)
Reports excluded: (n = 472) -Not qualitative (n = 83) -Children aged ≤7 years not included (n = 213) -No clear evidence of methods used for children ≤7 years (n = 159) -Not subjective health (n = 3) -Children ≤7 years cannot self-report with the PROM (n = 2) -Conference abstract (n = 8) -Dissertation/thesis (n = 2) -Outcome measure (n = 1) -Overview paper (n = 1)
Reports assessed for eligibility (n = 50)
Reports excluded: (n = 48) -Not qualitative (n = 6) -No children ≤7 included (n = 6) -No clear evidence of methods used for children ≤7 years (n = 34) -Not subjective health (n = 2)
Studies included in review (n = 19)

Figure 3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Frequency of studies
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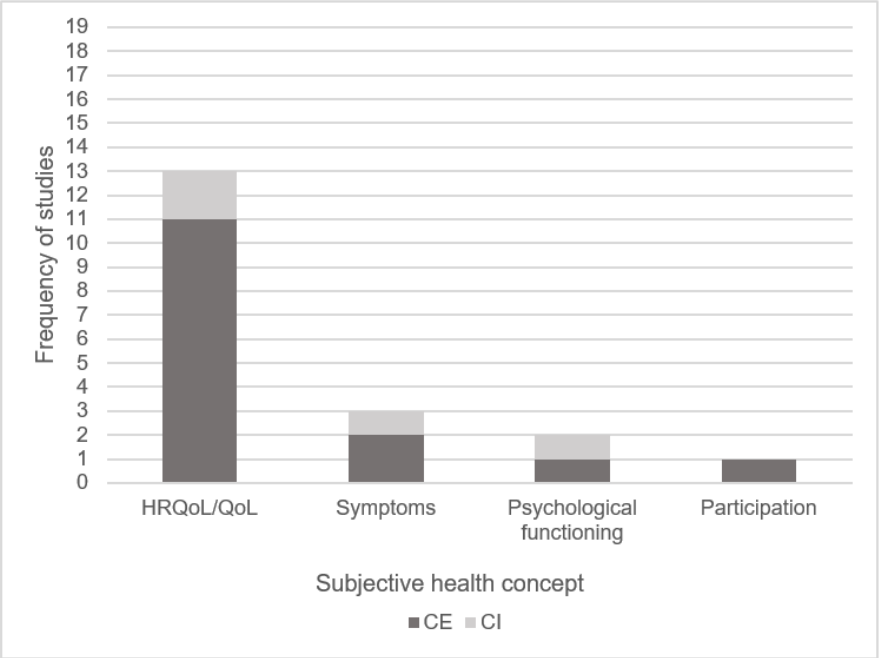
5 years
6 years
Youngest age (years) from which children were involved in studies

Figure 4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Frequency of studies
HRQoL/QoL
Symptoms
Psychological functioning
Participation
Subjective health concept
CE
CI