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Establishing content validity of LIMB-Q Kids: a new patient-reported outcome measure for lower limb deformities

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Abstract

Purpose To establish the content validity of LIMB-Q Kids, a new patient-reported outcome measure (PROM) for children with lower limb deformities. This study focused on three key components of content validity, i.e., comprehension, comprehensiveness, and relevance.

Methods Cognitive debriefing interviews (CDIs) with children with lower limb deformities were performed, and expert input from parents and clinicians directly involved in the care of children with lower limb deformities was obtained. CDIs were conducted with children from sites in Australia, Canada, and the USA. All interviews were recorded and transcribed verbatim. Analyses were conducted using the reparative approach, which involved close examination of the transcribed interviews and summarising edits after each interview.

Results Forty patients, experts, and parents reviewed LIMB-Q Kids and provided feedback. At the start, LIMB-Q Kids had 10 scales and 124 total items. Five rounds of input was obtained from children, parents, and experts. Overall, 37 new items were added. Thirty-three of the new items were included to measure symptoms experienced in different parts of the legs. Final version of LIMB-Q Kids had 11 scales and 159 items.

Conclusion This study established content validity of the LIMB-Q Kids, a new PROM for children with lower limb deformities. An international field-test study is underway. Translation and cultural adaptation are underway for sites where English is not the first language. Scoring algorithms will be developed, following which the scale could be used to inform clinical practice and research.

Keywords Content validity · Children · Patient-reported outcomes · Lower limb deformities · Qualitative

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Introduction

Lower limb deformities describe a range of congenital and acquired conditions primarily including leg length discrepancy, lower limb deficiency, and associated angular and rotational deformities [1]. Depending on the type of lower limb deformity, treatment options vary from non-surgical approaches to complex surgical procedures. Non-surgical approaches may include the use of adaptive devices, prostheses, orthotics, and shoe lifts. Surgical approaches may include reconstructive or ablative (amputation) surgery combined with the use of prostheses and orthotics. Commonly used procedures for lengthening and deformity correction involve the use of an internal nail or an external fixator. Amputation is a non-reversible surgical procedure that is followed by fitting of a prosthesis. Each surgical approach has advantages and disadvantages. At present, there is an insufficient evidence to support one treatment over the other [2–4]. However, it is important for future research that compares amputation with limb reconstruction using more sophisticated reconstructive procedures that the patient perspective of outcomes is measured using condition-specific Patient-Reported outcome measures (PROMs). Indeed, the complex treatment procedures that children with lower limb deformities undergo along with the reduced physical and social participation can have a considerable effect on their health-related quality of life (HRQL) [5–7].

There is currently no rigorously developed, internationally applicable condition-specific PROM to assess HRQL of children with lower limb deformities [8]. Our team developed a new internationally applicable PROM called LIMB-Q Kids following the international guidelines for PROM development [9]. Concept elicitation involved a systematic review of literature [7] and an international qualitative study [9]. A conceptual framework of concepts most important to children with lower limb deformities was developed. This qualitative study was followed by item generation and scale development, which was guided by the qualitative interviews with the target patient population from different countries and the conceptual framework [10]. For each of the themes included in the conceptual framework, the codes developed during the qualitative analysis of interviews with the children were reviewed, and items were written to cover the concept of each code, which created an exhaustive list of items.

The items were made short and concise to reduce the demand on verbal memory for children [11, 12]. In developing draft items and scales, the wording used by the patients was used as much as possible to ensure that the items resonate well with the experiences of the children with lower limb deformities. The language of the items was kept simple and straightforward. Any colloquial terminology, complex, ambiguous, and double-barrelled items were avoided. Positive or neutral wording of the items was used whenever possible. Also, if a concept overlapped with another PROM developed, cognitively tested and validated by our team for children (e.g., CLEFT-Q, a PROM for children and young adults with cleft lip and palate), rather than creating new wording for that concept already validated items were used [13, 14]. The target patient population was 8–18 years of age. Most items were written at or below grade 3 Flesch–Kincaid (F–K) reading level [15–17]. Likert-type scale with four-labeled response options with mutually exclusive, conceptually exhaustive categories was used, as suggested in the literature [18]. A 1-week recall period was chosen for all scales except the Appearance Scale, whereby the patients were asked about how much they like the appearance of their leg now.

Once items and scales were developed, we established their content validity. Content validity is the empiric evidence that demonstrates that the items and domains of a measurement instrument are appropriate and comprehensive relative to its intended measurement concept, population, and use [19–21]. Guidance for establishing content validity of PROMs is outlined by the COnsensus-based Standards for the selection of health Measurement Instruments (COSMIN) and The Professional Society for Health Economics and Outcomes Research (ISPOR) [21–24].

The aim of this study was to establish the content validity of LIMB-Q Kids by conducting cognitive debriefing interviews (CDI) with children with lower limb deformities and obtaining expert input from parents and clinicians directly involved in the care of children with lower limb deformities.

The objectives of CDIs were to assess a respondent's comprehension of the items with respect to their intended meaning, comprehensiveness of the content included in the PROM, and relevance of the items to the participants. This step is important for understanding a child's perspective, thought patterns and vocabulary, and identification of any format or wording problems or difficulties with the instructions, item stems, recall period, or response options [23, 25, 26]. CDIs can also be useful in identifying any additional items that have not been developed based on the initial concept elicitation phase. Using children as content validity experts to provide feedback on self-report instruments for children is a critical step in developing psychometrically sound measures for children [27].

Methods

Individual semi-structured interviews were conducted with children with lower limb deformities from sites in Australia, Canada, and the USA. Ethics approval was obtained at the

primary study site (CW15-0215/H15-00514) and at each of the participating sites. Written informed consent was obtained from the parents and assent was obtained from the children. Expert input was obtained at the same time from the clinicians and allied healthcare professionals at each of the participating sites. Version 1 of LIMB-Q Kids used for CDIs and expert feedback consisted of the following 10 independently functioning scales, i.e., Physical Function, Leg Symptoms, Hip Symptoms, Knee Symptoms, Foot Symptoms, Leg-Related Distress, Appearance, School Function, Social Function, and Psychological Function. Three key concepts of content validity were evaluated: comprehension, comprehensiveness, and relevance.

Sampling

Purposive sampling was used to recruit a maximum variation sample of children with lower limb deformities that included patients who varied by type of lower limb deformities, treatment type, stage of treatment, age, and sex. Children aged 8 to 18 years with a confirmed diagnosis of a lower limb deformity who could speak and read English were eligible. Patients with lower limb deformities along with other medical conditions which may have a confounding effect on their HRQL, and conditions such as cognitive or developmental delay affecting their ability to write and communicate, were excluded from this study. Isolated deformities of hip and foot were also excluded.

Recruitment

Recruitment was done by the participating sites in consultation with the primary study site (senior author's site). All interviews were conducted by one interviewer (HC). Interviews were done either in-person or virtually through Zoom.

Interviews with children

The interview process followed the approach suggested by Willis [28, 29] and used a semi-structured interview guide (found in supplementary files, pages 2–5). At the beginning of the interview, children were made aware of their role as critical reviewers of the items rather than just responding to the items [30]. A 'Think Aloud' protocol along with verbal probing was used [28]. Children were given the instructions to think-aloud while completing the items. Specific verbal probes such as asking the respondent to reformulate an item or to define some of the key terms in their own words were used [31]. During the interviews, children were asked to comment on the ease of completion, appropriateness of the instructions, recall period, items, and response options. Children were then asked to explain the reasons for choosing their answer for each item. This method of interviewing allowed us to query a participant's understanding of each item and their interpretation of the instructions and response options. If children identified certain items or words as hard to understand, or the interviewer noted any hesitation from the children while reading a specific word or item, the child was asked to suggest alternate wording. To assess comprehensiveness, at the end of the interview, children were asked to tell us anything about their leg that was important to them but was not included in the items reviewed. To assess relevance, children were asked about the importance of the items to them, and whether they can relate the items to their own experiences of having a lower limb deformity.

Expert and parent interviews

LIMB-Q Kids was shown to experts at each of the participating sites including orthopedic surgeons, nurses, physiotherapists, occupational therapists, psychologists, and prosthetists. Expert interviews were individual one-on-one interviews either in-person or virtually through Zoom (interview guide found in supplementary files, page 6). The purpose of seeking feedback from clinical experts was to ensure that all clinically relevant content was included. Experts were asked to comment on the comprehensibility of the instructions, items, and response options. Specifically, they were asked to identify items that they thought would be difficult for their patients to understand. In addition, they were asked to suggest alternate wording for any items they identified as including words 'not commonly used' by their patients. In addition, they were asked to identify any items that they thought were not relevant for their patient population. The experts were also asked to suggest any new items relevant for this patient population. Finally, they were asked whether they thought the items would be able to measure change when administered before and after treatments that they provide. Parents volunteered to provide feedback on the items as well. The questions for parents were similar to the questions asked to the clinical experts, i.e., questions focused on comprehension, comprehensiveness, and relevance of items to this patient population.

Analysis

All interviews were recorded and transcribed verbatim. Analyses were conducted using the reparative approach representing the 'Inspect and Repair' model [29]. This approach included closely examining the transcribed interviews and summarising edits after each interview. CDIs and expert interviews were conducted in rounds. A summary of feedback from patients and experts was prepared at the end of each round and reviewed by three members of the research team, who made changes to the scales as needed (Table 3). The research team members included an orthopedic surgeon

with expertise in treating children with lower limb deformities (AC) and experts in PROM development and HRQL (AK, JK). In an iterative manner, LIMB-Q Kids was revised through feedback until content validity was achieved (no further edits were suggested).

Results

Forty patients, experts, and parents reviewed LIMB-Q Kids between December 2019 and January 2021 (Tables 1 and 2). Mean age of the sample of 17 children at the time of CDI was 13 years (range 8 to 17 years). Nineteen experts and 4 parents provided feedback on the draft of LIMB-Q Kids (Table 2).

At the start of the study, Limb-Q Kids included 10 scales and 124 total items. Five rounds of interviews were conducted with children, parents, and experts (Table 3). Table 4 shows the number of changes made after each round. Overall, 37 new items were added in over the course of 5 rounds. Thirty-three of the new items were included in symptom scales. Four new items were added to the Physical Function Scale. Examples of item revisions are provided in Table 5. The final version of LIMB-Q Kids consists of 11 scales with 159 items.

Table 1 Demographics and clinical characteristics of patients participating in cognitive debriefing interviews

Demographic and clinical variables	Number (%)
Country	
Australia	5 (29.4)
Canada	11 (64.7)
USA	1 (5.9)
Sex	
Male	10 (58.8)
Female	7 (41.2)
Age (years)	
8–10	4 (23.5)
11–13	6 (35.3)
14–18	7 (41.2)
Average (range)	13 (8 to 17)
Type of deformity	
Fibular hemimelia	6 (35.3)
Amputation	4 (23.5)
Hemi-hypertrophy	2 (11.8)
Ollier's disease	1 (5.9)
Spondylocostal dysostosis and Scoliosis	1 (5.9)
Posteromedial tibial bowing and leg length discrepancy	1 (5.9)
Congenital short femur	1 (5.9)
Leg length discrepancy and dysplastic lateral femoral condyle	1 (5.9)

Table 2 Characteristics of parents and experts who provided feedback

Speciality	Australia	Canada	Ethiopia	India	UK	USA	Total (%)
Orthopedic surgeon	1	1	1	1	2	3	9 (39)
Nurse practitioner	0	1	0	0	0	0	1 (4)
Physiotherapist	1	2	0	0	1	0	4 (17)
Occupational therapist	0	2	0	0	0	0	2 (9)
Psychologist	0	0	0	0	1	0	1 (4)
Prosthetist	0	1	0	0	1	0	2 (9)
Parent	0	4	0	0	0	0	4 (17)
Total	2 (9)	13 (57)	1 (4)	1 (4)	5 (22)	3 (13)	23 (100)

Table 3 Sequence of cognitive debriefing interviews and expert interviews

	Country	N
Round 1		
Patients	Canada	5
Experts	Canada	3
Consultation with an orthopedic surgeon and experts in PROM development and HRQL		
Round 2		
Patients	Australia	2
	Canada	5
	US	1
Experts	Australia	1
	UK	2
	US	2
Parents	Canada	1
Consultation with an orthopedic surgeon and experts in PROM development and HRQL		
Round 3		
Patients	Australia	2
Experts	Australia	1
	Canada	3
	Ethiopia	1
	India	1
	UK	2
	US	1
Consultation with an orthopedic surgeon and experts in PROM development and HRQL		
Round 4		
Patients	Canada	3
Experts	Canada	3
	UK	2
	US	2
Parents	Canada	3
Consultation with an orthopedic surgeon and experts in PROM development and HRQL		
Round 5		
Patients	Australia	1
Consultation with an orthopedic surgeon and experts in PROM development and HRQL		

Summary of CDIs and expert feedback

Version 1: Overall, 91% of the items remained unchanged after the first round of interviews. By the end of round 1, LIMB-Q Kids consisted of 129 items.

Version 2: Eighty two percent of items remained unchanged after round 2 of interviews. A new Ankle Symptoms scale, with 8 items, was added after round 2 as suggested by the experts. By the end of round 2, LIMB-Q Kids consisted of 145 items.

Version 3: Ninety percent of the items remained unchanged after round 3. Seventeen items were added and 7 items were dropped. Out of the 17 items added, 15 were added to symptom scales. Fourteen out of 15 edits required during this round were minor changes to simplify wording and reduce the F–K grade level. At the end of round 3, LIMB-Q Kids consisted of 156 items.

Version 4: To ensure LIMB-Q Kids was relevant to amputations, round 4 included 3 patients with amputations, 3 parents of children with amputations, and 7 experts. Patients, parents, and experts only reviewed the Leg Symptoms Scale for any additional items that need to be added for children with amputations. Three items were added to the Leg Symptom Scale at this time.

Version 5: During round 5, the final version of LIMB-Q Kids had 159 items. No further changes were required based on patient feedback.

Table 4 Number of items retained, revised, dropped, and added during each round

Items	Round 1 Patients = 5 Experts = 3	Round 2 Patients = 8 Experts = 7 Parents = 1	Round 3 Patients = 2 Experts = 9	Round 4 Patients = 3 Experts = 7 Parents = 3	Round 5 Patients = 1
Physical Function					
Retained	27	23	30	32	32
Revised	1	5	9	0	0
Dropped	0	0	0	0	0
Added	0	2	2	0	0
Total	28	30	32	32	32
Leg Symptoms					
Retained	6	4	9	11	14
Revised	1	3	2	0	0
Dropped	0	0	0	0	0
Added	0	2	2	3	0
Total	7	9	11	14	14
Hip Symptoms					
Retained	2	5	7	10	10
Revised	3	2	2	0	0
Dropped	0	1	2	0	0
Added	3	2	3	0	0
Total	8	9	10	10	10
Knee Symptoms					
Retained	2	2	9	13	13
Revised	3	4	2	0	0
Dropped	0	1	0	0	0
Added	2	3	4	0	0
Total	7	9	13	13	13
Ankle Symptoms					
Retained	N/A		8	10	10
Revised			1	0	0
Dropped			0	0	0
Added			2	0	0
Total			10	10	10
Foot Symptoms					
Retained	4	4	5	9	9
Revised	2	2	1	0	0
Dropped	0	0	3	0	0
Added	0	2	4	0	0
Total	6	8	9	9	9
Leg-Related Distress					
Retained	14	13	13	13	14
Revised	1	2	1	0	0
Dropped	0	0	1	0	0
Added	0	0	0	0	0
Total	15	15	14	14	14
Appearance					
Retained	18	14	17	17	17
Revised	0	4	0	0	0
Dropped	0	0	1	0	0
Added	0	0	0	0	0
Total	18	18	17	17	17

Table 4 (continued)

Items	Round 1 Patients = 5 Experts = 3	Round 2 Patients = 8 Experts = 7 Parents = 1	Round 3 Patients = 2 Experts = 9	Round 4 Patients = 3 Experts = 7 Parents = 3	Round 5 Patients = 1
School Function					
Retained	17	15	16	17	17
Revised	0	1	1	0	0
Dropped	0	1	0	0	0
Added	0	1	0	0	0
Total	17	17	17	17	17
Social Function					
Retained	13	12	12	12	12
Revised	0	0	0	0	0
Dropped	0	1	0	0	0
Added	0	0	0	0	0
Total	13	12	12	12	12
Psychological Function					
Retained	10	10	10	10	10
Revised	0	0	0	0	0
Dropped	0	0	0	0	0
Added	0	0	0	0	0
Total	10	10	10	10	10

Note: Some experts from previous rounds were interviewed again in round 4 to review the Leg Symptoms Scale. These experts were only counted once in the total number of experts

Response options and recall period

All participants, including the youngest child who was 8 years old, found the response options and recall period easy to understand. Based on feedback from patients, parents, and experts, no changes were made to the response options and recall period for any of the scales throughout the course of the study.

Tables 5 and 6 show some quotes from interviews with patients and experts to support the edits to the items, instructions, and response options.

Physical Function Scale

This scale consists of items measuring the physical function required for day-to-day activities of daily life, sports, and recreation. Minor edits were made to instructions to avoid any confusion and introduce easier words based on the CDIs with the children, expert interviews, and feedback from parents (Table 5). This scale had 28 items at the beginning of the CDIs and 32 items in its final version. Overall, 16 out of 28 initial items remained unchanged throughout all five rounds.

Leg Symptoms Scale

This scale consists of items measuring symptoms in the leg. Based on the feedback for instructions for the Physical Function Scale, similar edits were made for instructions for all symptom scales (Leg symptoms, hip symptoms, knee symptoms, ankle symptoms, and foot symptoms). For example, instruction was changed from 'Do you have problems with both legs? If yes, please answer thinking of the leg that is worse' to 'These questions ask about the leg you are seeing the doctor or nurse about'. This edit was made as interviewed participants preferred not to call one of their leg as the worse leg. This scale had 7 items in version 1 and 14 items in its final version. Overall, 4 out of 7 items remained unchanged throughout all 3 rounds.

Hip Symptoms Scale

This scale consists of items measuring symptoms in the hip. This scale had 5 items in version 1 and 10 items in its final version. Three items were revised after round 1, 2 after round 2, and no items were revised after round 3 of interviews and expert input.

Knee Symptoms Scale

This scale consists of items measuring symptoms in the knee. This scale had 5 items in version 1 and 13 items in its final version.

Table 5 Examples of edits and supporting quotes from participants

Scale	Initial item	Final version of item	Participant quotes supporting edits
Physical Function	..lift your leg (e.g., to kick a ball)	..lift one leg while you stand on the other	*Patient:* If it was just lift your leg everybody would understand, like can you just lift your leg? If you're asking somebody of younger ages it would kinda be uh kick a ball it would be like two things so I think if you just left it as lift your leg they would more understand it. (12-year-old)
			Patient: Just lifting my leg yeah but kicking a ball to me implies force like bringing my leg back and then swinging it forward a little bit.(16-year-old)
Physical Function	..climb (e.g., up a ladder)	..climb (such as up a ladder)	*Patient:* What does eg stand for? (8-year-old)
Physical Function	..sit cross-legged	..sit cross-legged on the floor	*Patient:* and my, I'm thinking of sitting cross legged on like just the ground anywhere probably, so I personally right now can sit cross legged on my bed because my bed is squishy but I can't sit cross legged on a hard surface or floor. (16-year-old)
Leg Symptoms	My leg is puffy or swollen (bigger than normal)	My leg is swollen or puffy	*Patient:*..you might want to clarify if this is asking compared to the other one or if it has like increased in swollenness. I think that would be a good thing to put in a bracket, or just write my leg is more swollen. (17-year-old) *Patient*: My leg is swollen. What does swollen mean? I think we should call puffy instead of swollen. (8- year-old)
Hip Symptoms	My hip locks (gets stuck)	My hip gets stuck (not able to move it)	*Patient:* My hip locks… gets stuck. What I understand from this is like I guess also movement you can feel your hip like lock likes get stuck in this area or whatever. (12-year-old)
			Patient:…um like locking, like you can't move it, like it temporarily gets stuck. (age 17-year-old)
Knee Symptoms	My knee gives way when I move (feels wobbly)	My knee feels wobbly (like it is going to give out)	*Patient:* Um, I don't know, I am not sure if a younger kid would understand gives way that might be hard to understand, so that might be easier for them to understand like my knee feels wobbly. (17-year-old)
			Patient: My knee gives way when I move, meaning feeling wobbly. *Interviewer*: so what do you understand with this question? *Patient*: it's like when you're walking and your knee suddenly turns wobbly. (9-year-old)
Foot Symptoms	My foot hurts when I walk	(1) My foot hurts when I walk with my shoes on; (2) My foot hurts when I walk bare foot (no shoes on my feet)	*Expert:*.. when I'm talking with kids with feet issues, I would say, "at the end of the day, do you get pain in these specific areas?" Or "do you have to take your shoes off during the day to have a rest from your foot?" Or "do you have to," you know, "stop playing sports halfway through a game because your feet are rubbing?"

Table 5 (continued)

Scale	Initial item	Final version of item	Participant quotes supporting edits
Leg-related distress	I cover up or hide my leg when I go out	I hide my leg when I go out	*Patient:* I'm not really sure because I wear pants which is technically covering your legs but covering or hiding your legs would be like if you were wearing shorts, like if you had to be wearing shorts and maybe putting your leg behind your other leg, that to me would be hiding or covering up but the fact that I wear pants on a regular basis, I don't know if I would consider that hiding or covering my leg just by wearing pants. (16-year-old)
Leg-related distress	I avoid wearing shorts or skirts	I avoid wearing shorts or skirts that show my leg	*Patient:* I just don't understand why someone would not wear shorts or skirts. (13-year-old)
Leg-related distress	I dislike how I look when I walk (e.g., limp)	I dislike how I look when I walk	*Patient:* um I guess I look a little misaligned but don't necessarily think I limp I just look a little funky. (16-year-old)
Appearance	..how well your hips match each other (straight or tilted)	..how well your hips match each other (straight or uneven)?	*Patient:* How- how much do you like how well your hips match each other. What does tilted mean? (8-year-old)
Appearance	..how straight your leg looks (not crooked or lopsided)	..how straight your leg looks?	*Patient:* well it's not crooked or lopsided. It looks more different from other people's so I don't really like it that much. (9-year-old)
Appearance	How much do you like the size of your leg (not too skinny)	How much do you like the size of your leg	*Parent:* … get rid of "not too skinny". I don't like the word skinny. Girls shouldn't be looking at that word "skinny"
Scale	Initial instructions	Final version of instructions	Participant quotes supporting edits
Physical Function	Do you have problems with both legs? If yes, please answer thinking of the leg that is worse	These questions ask about the leg you are seeing the doctor or nurse about	*Patient:* I don't want to say worse leg, because it isn't worse but like the one that had a little more problems. (12-year-old patient) *Parent:* It's like if we frame their leg that is theirs and they're answering questions about their leg and they're hearing that their leg is a problem. It's not the kindest way that we can describe their physical challenge
Physical Function	If you use a special shoe, shoe-lift, splint, prosthesis and any other special device or have a frame on, please answer thinking about how your leg feels with those devices on	If you wear a special shoe, shoe-lift or brace or have an artificial leg (prosthetic leg), please answer the questions thinking of when you have them on'	*Patient:* Maybe instead of prosthesis because it's definitely harder to say. Maybe like prosthetic or maybe artificial leg. Yeah, either of those would be better. I feel like that one's kind of easier to understand. (13-year-old patient)

Table 6 Quotes from interviews with experts

Expert	Participant quotes
Experts—response options	*Physiotherapist:* I think generally from a follow up perspective we tend to ask more about um… how often, how much, more than how much. Because like um… the how often kind of gives you a better idea of how limited they would be in their function. You know because, is it like… is it like all the time then they're probably more likely to have more functional impairment
	Surgeon: …I think they will relate more closely to frequency. Yeah, I find that for example when patients give me back a PROM, I usually do it on paper just because it's easier….., these types of questions can cause some confusion like sort of question on it, like I'm not sure I understand, or they'll draw an arrow and make it sort of in between two of them because it's much but it's not very much so yeah, definitely frequency would be more objective from a patients point of view
Experts—items	*Surgeon:* So… let's see… get out of bed… sit in a chair – I don't – get up from a chair. These are all – I mean, these are all things that I've had people complain of, so this is good
	Surgeon: It's a pretty – a pretty um, detailed list. Overall, it's very comprehensive. Surgeon: I think these are great questions because they cover the gamut of the symptoms that I would be asking about and usually the patients come in with pain, or having weakness, or some neurologic type symptoms and pain at rest is obviously an important one too because it may point to other things so I think these are all very good questions and they're all understandable too, you know putting the word shaky, cannot feel it, pins and needles, I think those are all very helpful
Experts—responsiveness	*Interviewer:* Do you think like, these items or some of these items are able to measure change? *Surgeon:* Yeah, yeah I do think that there is some, you know, certain things like "bend your knees," sometimes that's a surgical goal right? Um… yeah, some things so those are like range of motion things, "walk without limping," we're often times trying to fix a limp… um, speed and distance is certainly the way a lot of people um… gauge whether their surgery has helped, um, just in a very informal way, so having it like more quantified as you have here would be helpful
	Surgeon: I think, this reasonably covers most areas possible which might show a change for us in our final analysis if we do surgery or whatever treatment you do and you want to come back in two years' time and ask the same questions, yeah
	Interviewer: Thinking about the social life and the school scale, do you think you can measure change with these items. *Surgeon:* I would think so yeah, because I've observed myself with many of my patients, I keep asking them as well, actually, many of these questions I ask especially at the end of treatment. I ask many of them what they think of themselves and what their friends think
Expert—use in clinical practice	*Interviewer:* Do you see yourself using these scales? *Surgeon:* oh yeah, definitely, yeah one of the complaints that I would have about a lot of the scales that I use are that they don't capture a global picture about how this affects somebody. I really appreciate those sections in terms of quality of life and things like that because those are the ones that often children find the most like you said, the most problems they have is the distress they have because of whatever physical problem they have and it's interesting to see both like in terms of how their functional sections look in terms of and how their quality of life section looks because it's very commonly their function isn't as impacted as, maybe their score isn't very low in that section but their quality of life is terribly low and you know they worry about. So definitely for me, that's one of the big advantages I think of having a deformity specific questionnaire, this stuff is as important as the functional stuff
	Physiotherapist: I think it's something very useful. It's a really interesting tool and hopefully it will change our practice too. For us to know what's going on in their life while they are going through a treatment and before

Ankle Symptoms Scale

This scale consists of items measuring symptoms in the ankle. As suggested by the clinical experts, a separate scale for ankle symptoms was added after round 2 of interviews and expert feedback. Instructions for this scale were kept similar to other symptom scales. This scale had 10 items in its final version. The 8 items added for this scale were similar in wording to the other symptom scales. These items were tested in round 3. Two new items were added to the ankle symptoms scale based on the interviews and expert feedback in round 3.

Foot Symptoms Scale

This scale consists of items measuring symptoms in the foot. This scale had 6 items in version 1 and 9 items in its final version. Two items were revised after round 1, 2 after round 2, and no items were revised after round 3 of interviews and expert input.

Leg-Related Distress Scale

This scale is composed of items measuring the distress the children feel due to their leg.

The items ask about how the children feel about their leg. In round 1, 3/5 participants were not clear that it was about how they feel about their leg. One participant suggested underlining 'you feel'. As a result, the instructions were edited to underline 'you feel' to emphasize that it is about how the person feels rather than how the leg feels (as in the symptoms scales). This scale had 15 items in version 1 and 14 items in its final version. The item 'I feel unhappy about the scars on my leg' was included in rounds 1 and 2 of CDIs and expert input. This item was found relevant only to patients who had scars. Hence, this item was removed from this scale. It was kept as a stand-alone item to be asked only of patients who had scars due to their surgeries.

Appearance Scale

This scale consists of items asking about the appearance-related concerns of the children. After round 2 of interviews and expert feedback, the instruction 'Do your legs look different from each other? If yes, please answer thinking of the leg that you like the least' was edited to 'These questions ask about the leg you are seeing the doctor or nurse about.' This scale had 18 items in version 1 and 17 items in its final version. Thirteen out of 18 items remained unchanged during all five rounds of interviews and expert input. An item about the appearance of scars was included in rounds 1 and 2 of CDIs and expert input. This item was removed from this scale and kept as a stand-alone item that is only applicable to patients with scars.

Other scales

School function scale, social function scale, and the psychological function scale needed minimal edits. No edits were made to the instructions for either of these scales.

General expert feedback

Overall, the experts found the scales to be comprehensive and the items in each scale easy to understand and relevant to this patient population. The majority of the additional items suggested were for the symptom scales. Some quotes from expert interviews are presented in Table 6.

When specifically asked whether they think these items will be able to measure change, all experts thought most of the items would be sensitive to measuring change when administered before and after treatment. Experts were asked whether they would potentially use the LIMB-Q Kids in their clinical practice. Most of them were interested in using LIMB-Q Kids when finalized and validated.

Readability

Mean F–K readability for the 159 items was 1.04 (range 0–5.2) (Table 1 in Supplementary Material). The readability scores for 90% of items were at or below grade 3 F–K reading level. The 10% items that were over grade 3 F–K reading level were still understood well by the patients interviewed during the cognitive debriefing interviews.

Discussion

CDIs with an international sample of patients and feedback from clinical and methodological experts established the content validity of LIMB-Q Kids. We were able to confirm suitability of the response options, and refine the instructions and items for each scale. Feedback from parents and a diverse group of experts involved in the care of patients with lower limb deformities helped to establish comprehensiveness and ensure the clinical relevance of the item forming each scale to this patient population.

Following the COSMIN guidelines for establishing content validity of PROMs, we focused on three key components of content validity, i.e., comprehension, comprehensiveness, and relevance. Our aim was to develop items applicable to and understood by patients with lower limb deformities between the ages of 8 and 18 years. After making relatively minor edits, the items and instructions were understood by all interviewed patients. The majority of items were at F–K grade level of 3 or lower. A low-grade reading level is crucial for PROMs for children. The majority of the children did not suggest any new items and found the items relevant to their experience with their lower limb deformities. Overall, the experts found the scales to be comprehensive and covering areas relevant to this patient population. Most of the new items suggested by experts were added to the symptom scales.

Based on our systematic review in 2016 and our recent literature search, there is still a lack of a rigorously developed and internationally applicable PROM for children with lower limb deformities [8]. Most experts interviewed in this study indicated their interest in using LIMB-Q Kids in their practice due to the lack of such a comprehensive and rigorously developed condition-specific tool. In the absence of condition-specific PROMs for this patient population, previous studies have used generic outcome measurement tools or parent-reported measurement instruments [8]. A paper published in 2019 discusses the use of a 5-item Lower Limb Satisfaction Questionnaire for children with lower limb deformities [3]. This child version was developed from a parent-reported version of the Limb-Lengthening Satisfaction Questionnaire, which was adapted from a parent questionnaire for assessing prosthesis satisfaction in children

with limb deficiencies only (CAPP-PSI) [32]. However, there are no available reports on the detailed development and validation of the child version of the Lower Limb Satisfaction Questionnaire. Recently, a new questionnaire for gait outcome assessment for children with lower limb differences has been developed [33]. However, the development and initial validation studies were only conducted in developed countries including USA, Australia, and Canada. There has been no involvement from patients and experts from low- and middle-income countries during the development and initial validation of this new questionnaire. Hence, the development of LIMB-Q Kids by engaging patients and clinical experts from low- and middle-income countries makes it potentially more relevant and applicable to children and adolescents with lower limb deformities internationally. Modified version of Scoliosis Research Society (SRS) outcome measure, Limb Deformity Modified SRS (LD-SRS) has been used for adults with lower limb deformities [34–36]. Hence, the need for a PROM that is developed by engaging the target patient population during concept elicitation and content validation still exists.

The inclusion of an international sample of patients and experts is a strength of this study. Clinical experts from various international sites, directly involved in the care of children with lower limb deformities, were consulted for their feedback. The study team consisted of experts in PROM development and HRQL studies. All CDIs with patients and interviews with experts were conducted by one interviewer to ensure consistent methodology. The detailed one-on-one interviews with the patients and clinical experts provided rich feedback for the items included in each scale.

A limitation of our study is that we were not able to conduct CDIs with children from other low-income and lower-income countries as part of this content validation study. We were able to include children from Ethiopia and India during the initial qualitative phase (10). However, the inclusion of clinical experts from some of these countries (Ethiopia and India) was useful to ensure that the items reflected the perspective of children in these countries. The authors plan to include more low-income and lower-income countries in the translation and cultural adaptation phase and the international field test.

Conclusions

This study demonstrated content validity of the scales included in LIMB-Q Kids, a new PROM for children with lower limb deformities. An international field-test study is now underway. Data will be collected from a large sample of children with lower limb conditions. Psychometric analysis will be performed using a modern approach (i.e., Rasch Measurement Theory) to explore each scale's psychometric performance and to develop the scoring algorithm for each scale [37]. Items that do not perform well will be dropped. Importantly, given the modern psychometric approach, each LIMB-Q Kids scale is independently functioning, which means that researchers and clinicians will be able to choose a subset of scales depending on the research question or clinical concern. This modular approach to PROM development makes it possible to reduce respondent burden by keeping the length of a PROM manageable for children [38]. Translation and cultural adaptation is being conducted for sites where English is not the first language.

When fully developed and validated, LIMB-Q Kids can be used to assess HRQL of children with lower limb deformities. In the long term, LIMB-Q Kids could be used by healthcare professionals to look at the effectiveness of interventions in terms of improvements in leg appearance, function, symptoms, and overall HRQL. This process will eventually help develop or modify interventions/treatment plans, health care services, and target care, to achieve a better HRQL for children with lower limb deformities. LIMB-Q Kids can also help future patients in making important decisions about treatment choices, by comparing HRQL between different interventions (such as amputation versus reconstruction) and comparing HRQL pre- and post-intervention.

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Data availability Any further data from this study will be available upon request from the first author.

Code availability N/A.

Declarations

Conflict of interest There are no financial or non-financial interests to disclose.

Ethical approval Ethics approval was obtained at the primary study site (CW15-0215/H15-00514) and at each of the participating sites. This study adhered to the principles of the Declaration of Helsinki.

Consent to participate A written informed consent was obtained from the parents and assent was obtained from the children by the participating sites.

Consent to publish Publication of study results in academic journals was included in the consent to participate for this study.

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