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SYMPOSIUM: PSYCHOSOCIAL ASPECTS OF MUSCULOSKELETAL ILLNESS

Do Patient- and Parent-reported Outcomes Measures for Children With Congenital Hand Differences Capture WHO-ICF Domains?

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Abstract

Background Patient- and parent-reported outcome measures (PROMs) are increasingly used to evaluate the effectiveness of surgery for congenital hand differences (CHDs). Knowledge of an existing outcome measure's ability to assess self-reported health, including psychosocial aspects, can inform the future development and application of PROMs for CHD. However, the extent to which measures used among children with CHD align with common, accepted metrics of self-reported disability remains unexplored.

Questions/purposes We reviewed studies that used PROMs to evaluate surgery for CHD to determine (1) the number of World Health Organization-International Classification of Functioning, Disability and Health (WHO-ICF) domains covered by existing PROMs; (2) the proportion of studies that used PROMs specifically validated among children with CHD; and (3) the proportion of PROMs that targets patients and/or parents.

Methods We performed a comprehensive review of the literature through a bibliographic search of MEDLINE®, PubMed, and EMBASE from January 1966 to December 2014 to identify articles related to patient outcomes and surgery for CHD. We evaluated the 42 studies that used PROMs to identify the number and type of WHO-ICF domains captured by existing PROMs for CHD and the proportion of studies that use PROMs validated for use among children with CHD. The most common instruments used to measure patient- and parent-reported outcomes after reconstruction for CHD included the Prosthetic Upper Extremity Functional Index (PUFI), Disabilities of the Arm, Shoulder, and Hand questionnaire, Childhood Experience Questionnaire, and Pediatric Quality of Life Inventory.

Results Current PROMs that have been used for CHD covered a mean of 1.3 WHO-ICF domains (SD ± 1.3). Only the Child Behavior Checklist and the Piers-Harris Children's Self-Concept Scale captured all ICF domains (body functions and structures, activity, participation, and environmental factors). The PUFI, the only PROM validated specifically for children with congenital longitudinal and transverse deficiency, was used in only four of 42 studies. Only 13 of the 42 studies assessed patient-reported outcomes, whereas five assessed both patient- and parent-reported outcomes.

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Conclusions The PROMs used to assess patients after CHD surgery do not evaluate all WHO-ICF domains (ie, body structure, body function, environmental factors, and activity and participation) and generally are not validated for children with CHD. Given the psychological and sociological aspects of CHD illness, a PROM that encompasses all components of the biopsychosocial model of illness and validated in children with CHD is desirable.
Level of Evidence Level III, therapeutic study.

Introduction

Congenital hand differences (CHDs) affect approximately one in every 600 newborns [59]. More than 10% of these children have partial or complete absence of the hand and functional losses are substantial [9]. Furthermore, children and their parents invariably experience considerable psychological stress from the appearance of the involved limb. Previous studies demonstrate that parents of children with CHD frequently mourn the loss of their expectations for their unborn child, and children with CHD have difficulty coping with the functional and aesthetic manifestations of these conditions [12, 30]. In this context, patient- and parent-reported outcome measures (PROMs) that can accurately and efficiently capture these experiences could provide important insight into the effect of CHDs on a child's psychosocial functioning and development.

Many large series have reported substantial improvement in objective outcomes after surgery for CHD [29, 36, 48, 53, 54, 61–63]. However, the ultimate measure of success depends on more than the traditional measures of strength, sensibility, ROM, and time to task completion [1, 65]. In 2001, the World Health Organization developed the International Classification of Functioning, Disability, and Health (WHO-ICF) by which self-reported health can be considered and classified and consists of four domains: (1) body structure; (2) body function; (3) environmental factors; and (4) activity and participation [28]. Its purpose is to create an integrated biopsychosocial model of health status that accounts for environmental, sociodemographic, and psychological factors and to standardize descriptions of health and health-related status [79]. The biopsychosocial concept is a scientific model developed in the 1970s to account for the complex interplay of both medical and social factors that affect one's health status [27]. The traditional biomedical model may be an effective way of measuring patient-reported outcomes but is limited by an inability to capture factors beyond the patient or disease process. Furthermore, because the biopsychosocial model categorizes self-reported health into discrete components, the ICF provides a clarifying framework for systematically reviewing the content of patient-reported outcome measures [51]. The use of a specific PROM can be assessed by examining the correlation between survey items and domains of this conceptual model [70].

The importance of patient and parent perspectives regarding disability and health status have been increasingly recognized, and PROMs are commonly used to evaluate the effectiveness of surgery for many conditions [6, 13, 18, 47, 60, 68]. However, prior research suggests that children's experiences are not consistently elicited during clinical care, and providers more often engage parents during routine encounters [74, 75]. In turn, children may report feeling unheard and disconnected from providers [17, 32, 69] and, as a result, parent and child perceptions of outcomes, risks, and satisfaction are often discordant [24, 39, 57]. Therefore, understanding the extent to which child and parent perception of outcomes are captured is critical to develop effective and appropriate treatment plans.

To date, the extent to which PROMs are applied toward children with CHD is unclear, and the degree to which specific instruments accurately capture psychosocial functioning and disability is unknown. Understanding the ability of existing PROMs to assess such outcomes is critical to refine and improve these methods for future work. The purpose of this study is to determine (1) the number of WHO-ICF domains covered by existing PROMs; (2) the proportion of studies that used PROMs specifically validated among children with CHD; and (3) the proportion of PROMs that target patients and/or parents.

Search Strategy and Criteria

We performed a bibliographic search of MEDLINE®, PubMed, and EMBASE from January 1966 to December 2014 to identify articles related to patient outcomes after surgery for CHD. We used the phrases and keywords "brachydactyly", "brachysyndactyly", "camptodactyly", "cleft hand", "clinodactyly", "congenital hand anomalies", "congenital hand differences", "congenital hand abnormalities", "congenital clasped thumb", "finger malformation", "hand malformation", "hand anomaly", "hand deformities, congenital", "mirror hand", "pediatric hand anomalies", "pediatric hand conditions", "pediatric hand deformities", "polydactyly", "radial deficiency", "radial longitudinal deficiency", "radial dysplasia", "syndactyly", "thumb deficiency", "thumb duplication", "thumb hypoplasia", and "thumb malformation". We conducted a title and abstract search to identify appropriate articles using the following a priori criteria: original paper with primary patient outcomes; human subjects; English language publication published between January 1966 and

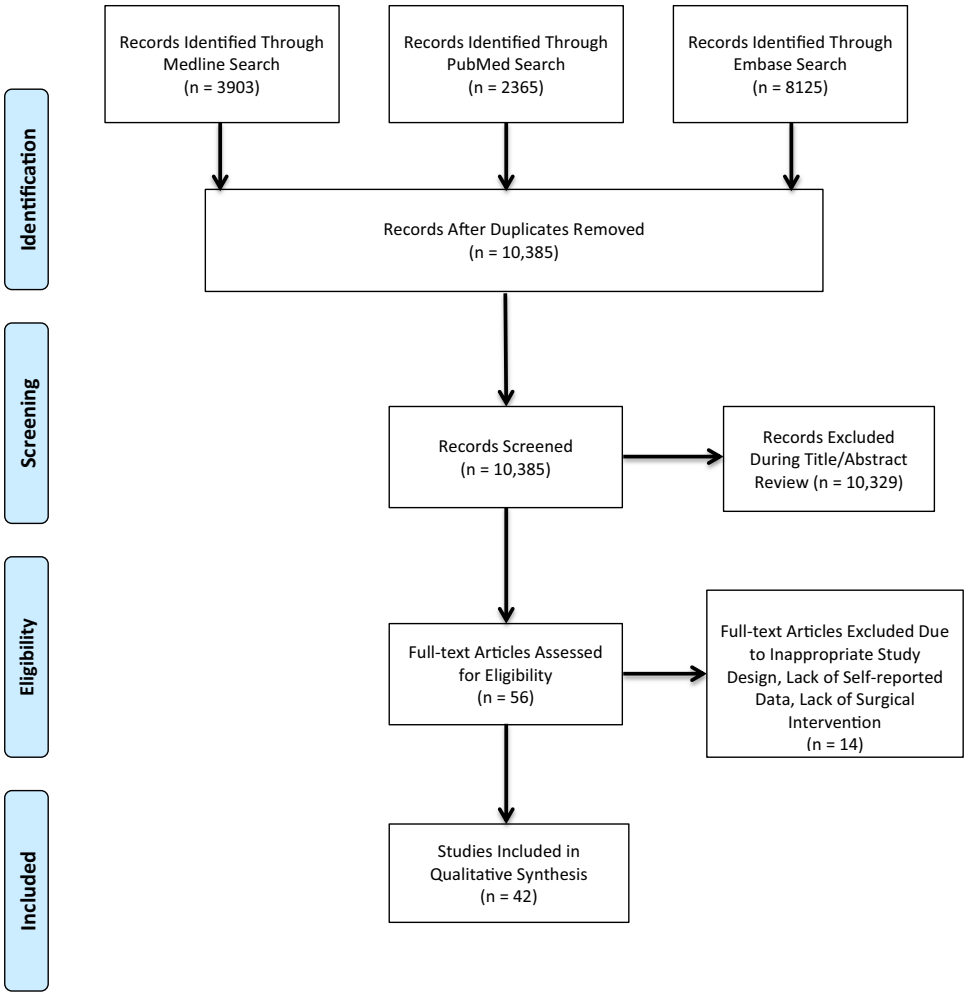

Fig. 1 This flowchart represents the primary database search including number of articles retrieved and excluded from review using PRISMA guidelines.

December 2014; treatment included surgery for CHDs; and patient age at the time of surgery 0 to 18 years old. Additionally, we performed a manual reference check of the retrieved articles to capture additional references not initially captured by the original search. We excluded articles from review if they reported investigations of nonoperatively managed congenital hand differences, lacked patient- or parent-reported outcomes, or were from non-English language citations.

Data Extraction and Analysis

For our search, we used the selection process outlined by the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) guidelines (Fig. 1). Our primary literature search identified 10,385 citations. Ultimately, 42 studies, including 1429 patients from 12 different countries, met inclusion criteria. Of these, 39 were cohort studies [2, 3, 5, 11, 12, 14–16, 19, 21–23, 26, 31, 33–35, 37, 38, 41, 44–46, 48, 50, 52, 56, 58, 62, 64, 66, 71–73, 76–78, 80, 82], two were cross-sectional studies [7, 43], and one was a case-control study [4].

After formal article review, two reviewers (JMA, RSB) independently extracted data from each study. These data included: study type, level of evidence, type of CHD, sample size, patient demographics, procedure performed, length of followup, domains and results of surgeon-generated ad hoc questionnaires, type, timing, and results of any patient- or parent-reported outcomes measure, clinical examination findings (when available), and ICF domain comparison for validated PROMs (Table 1).

Surgeon-generated ad hoc questionnaires were used in 36 studies. Most commonly, surgeon-generated questionnaires assessed satisfaction, appearance, function, social implications, psychosocial well-being, task performance, and pain. Objective outcomes measures most commonly evaluated included ROM, grip and pinch strength, tests of

Table 1. Content analysis of studies using patient- or parent-reported outcomes in patients with CHD

Title	Author	Study type	Level of Evidence	Sample size	Type of anomaly	Procedure(s) performed
Characteristics of Patients with Hypoplastic Thumb: A Prospective Study of 51 Patients with the Results of Surgical Treatment [2]	Abdel-Ghani and Amro	Cohort study	II	51 patients, 82 hands	Hypoplastic thumb	Opponensplasty and pollicization
A 5-Year Interval Evaluation of Function after Pollicization for Congenital Thumb Aplasia Using Multiple Outcome Measures [3]	Aliu et al.	Cohort study	II	5 patients, 7 hands	Hypoplastic thumb	Pollicization
Children with Surgically Corrected Hand Deformities and Upper Limb Deficiencies: Self-Concept and Psychological Well-Being [4]	Andersson et al.	Case- control	III	92 patients (79 treated surgically)	All congenital anomalies	Not reported
Duplicate Thumbs: A Survey of Results in Twenty Patients [5]	Andrew and Sykes	Cohort study	III	20 patients, 21 hands	Thumb duplication	Thumb reconstruction
Low Impact of Congenital Hand Differences on Health-Related Quality of Life [7]	Ardon et al.	Cross-sectional	III	116 patients (75 treated surgically), 155 hands	All congenital anomalies	Not reported
Psychological Aspects of Toe to Hand Transfer in Children [12]	Bellew and Kay	Cohort study	II	37 patients (32 congenital, 5 posttraumatic)	Multiple (most commonly symbrachydactyly)	Toe to hand transfer
Toe to Hand Transfer in Children: Ten Year Follow Up of Psychological Aspects [11]	Bellew et al.	Cohort study	II	25 patients (21 congenital, 4 posttraumatic)	Multiple (most commonly symbrachydactyly)	Toe to hand transfer
The Psychological Impact of Microvascular Free Toe Transfer for Children and Their Parents [14]	Bradbury et al.	Cohort study	II	14 patients	Not reported	Toe to hand transfer
Hand Function and Activity Performance of Children with Longitudinal Radial Deficiency [15]	Buffart et al.	Cohort study	III	20 patients, 27 hands	Radial longitudinal deficiency	Soft tissue distraction, centralization, pollicization, and opponensplasty
Outcome of Index Finger Pollicisation for the Congenital Absent or Severely Hypoplastic Thumb [16]	Ceulemans et al.	Cohort study	III	17 patients, 24 hands	Hypoplastic thumb	Pollicization
Pollicisation of the Index Finger: A 27 Year Follow Up Study [19]	Clark et al.	Cohort study	III	11 patients, 15 hands	Hypoplastic thumb	Pollicization
Outcome after Pollicization: Comparison of Patients with Mild and Severe Longitudinal Radial Deficiency [21]	de Kraker et al.	Cohort study	II	24 patients, 30 hands	Hypoplastic thumb	Pollicization
Comparison of Functional Outcome Scores in Radial Polydactyly [22]	Dijkman et al.	Cohort study	III	37 patients, 41 hands	Thumb duplication	Not reported
Perception of the Esthetic and Functional Outcomes of Duplicate Thumb Reconstruction [23]	Do et al.	Cohort study	III	31 patients, 33 thumbs	Thumb duplication	Thumb reconstruction

Table 1. continued

Title	Author	Study type	Level of Evidence	Sample size	Type of anomaly	Procedure(s) performed
Hand Function in Adults with Radial Longitudinal Deficiency [26]	Ekblom et al.	Cohort study	III	20 patients, 29 limbs	Radial longitudinal deficiency	Centralization, radialization, or pollicization
Radial Polydactyly: An Outcome Study [31]	Ganley and Lubahn	Cohort study	III	16 patients, 21 hands	Thumb duplication	Thumb reconstruction
Objective Features and Aesthetic Outcome of Pollicized Digits Compared With Normal Thumbs [34]	Goldfarb et al.	Cohort study	III	26 patients, 31 digits	Hypoplastic thumb	Pollicization
Thumb Size and Appearance Following Reconstruction of Radial Polydactyly [37]	Goldfarb et al.	Cohort study	III	26 patients, 31 digits	Thumb duplication	Thumb reconstruction
Central Ray Deficiency: Subjective and Objective Outcome of Cleft Reconstruction [33]	Goldfarb et al.	Cohort study	III	12 patients, 16 hands	Cleft hand	Cleft closure and reconstruction
Complex Syndactyly: Aesthetic and Objective Outcomes [38]	Goldfarb et al.	Cohort study	III	13 patients, 21 hands	Complex syndactyly	Reconstruction
Functional Outcome After Centralization for Radius Dysplasia [35]	Goldfarb et al.	Cohort study	III	21 patients, 25 wrists	Radial longitudinal deficiency	Centralization
Macrodactyly–Options and Outcomes [41]	Hardwicke et al.	Cohort study	III	20 upper limb (16 treated surgically)	Macrodactyly (upper and lower limbs)	Multiple (syndactyly release, nail bed/bone resection, osteotomy, arthrodesis, epiphysiodesis, ray amputation, debulking)
Long-term Functional Outcome of Patients with Longitudinal Radial Deficiency: Cross-Sectional Evaluation of Function, Activity and Participation [43]	Holtslag et al.	Cross-sectional study	III	17 patients, 31 arms	Radial longitudinal deficiency	Centralization
Outcome Measures of Microsurgical Toe Transfers for Reconstruction of Congenital and Traumatic Hand Anomalies [44]	Kaplan and Jones	Cohort study	II	15 patients, 20 toe transfers	Transverse failure of formation, symbrachydactyly, traumatic hand injuries	Toe to hand transfer
Toe to Hand Transfer in Children Part 2: Functional and Psychological Aspects [45]	Kay and Wiberg	Cohort study	II	37 patients, 37 hands	Congenital malformations, traumatic hand injuries	Toe to hand transfer
Pre-axial Polydactyly: Outcome of the Surgical Treatment [46]	Kemnitz and De	Cohort study	III	22 patients, 22 hands	Thumb duplication	Thumb reconstruction
Comparison of Surgical Treatment and Nonoperative Management for Radial Longitudinal Deficiency [48]	Kotwal et al.	Cohort study	III	239 patients, 309 hands treated surgically, (137 treated nonsurgically)	Radial longitudinal deficiency	Centralization or radialization
Long-Term Follow-Up of Surgical Treatment for Thumb Duplication [50]	Larsen and Nicolai	Cohort study	III	18 patients, 19 thumbs	Thumb duplication	Thumb reconstruction

Table 1. continued

Title	Author	Study type	Level of Evidence	Sample size	Type of anomaly	Procedure(s) performed
Long-Term Outcomes of Web Creep, Scar Quality, and Function After Simple Syndactyly Surgical Treatment [52]	Lumenta et al.	Cohort study	III	19 patients, 26 web spaces	Simple syndactyly	Surgical release with skin graft
The Preliminary Outcome of Syndactyly Management in Children with a New External Separation Device [56]	Mei et al.	Cohort study	II	12 patients, 12 hands	Syndactyly	External separation device followed by syndactyly release
Functional Outcomes of Children With Index Pollicizations for Thumb Deficiency [58]	Netscher et al.	Cohort study	II	18 patients, 22 hands	Hypoplastic thumb	Pollicization
Long-term Results of Surgical Treatment of Thumb Polydactyly [62]	Ogino et al.	Cohort study	II	105 patients, 113 thumbs	Thumb duplication	Thumb reconstruction
Factors Affecting Surgical Results of Wassel Type IV Thumb Duplications [64]	Patel et al.	Cohort study	III	41 patients, 42 thumbs	Thumb duplication	Thumb reconstruction
Functional Assessment After Pollicisation [66]	Roper and Turnbull	Cohort study	III	9 patients	Hypoplastic thumb	Pollicization
Functional Outcome for Children with Thumb Aplasia Undergoing Pollicization [71]	Staines et al.	Cohort study	II	10 patients, 12 thumbs	Hypoplastic thumb	Pollicization
Long-Term Outcomes Following Radial Polydactyly Reconstruction [72]	Stutz et al.	Cohort study	III	41 patients, 43 thumbs	Thumb duplication	Thumb reconstruction
Comparative Study of Outcomes between Pollicization and Microsurgical Second Toe- Metatarsal Bone Transfer for Congenital Radial Deficiency with Hypoplastic Thumb [73]	Tan and Tu	Cohort study	II	30 patients	Hypoplastic thumb	Pollicization or free second toe-metatarsal bone transfers
The Results of Pollicization for Congenital Thumb Hypoplasia [76]	Tonkin et al.	Cohort study	III	35 patients, 42 thumbs	Hypoplastic thumb	Pollicization
Outcome Analysis, Including Patient and Parental Satisfaction, Regarding Nonvascularized Free Toe Phalanx Transfer in Congenital Hand Deformities [77]	Unglaub et al.	Cohort study	III	20 patients, 56 toe transfers	Symbrachydactyly or constriction ring syndrome	Nonvascularized free toe phalanx transfers
Index Finger Pollicization in the Treatment of Congenitally Deficient Thumb [78]	Vekris et al.	Cohort study	III	18 patients, 21 hands	Hypoplastic thumb	Pollicization
Thumb Polydactyly: Clinical Outcome After Reconstruction [80]	Yen et al.	Cohort study	III	34 patients, 36 thumbs	Thumb duplication	Thumb reconstruction
Developing a Pollicization Outcomes Measure [82]	Zlotolow et al.	Cohort study	III	35 patients, 42 digits	Hypoplastic thumb	Pollicization

Table 1. continued

Title	Gender distribution	Average age at surgery (range)	Validated survey(s) used	Construct(s) measured	Country
Characteristics of Patients with Hypoplastic Thumb: A Prospective Study of 51 Patients with the Results of Surgical Treatment [2]	Male: 25 Female: 26	37 months (8 months to 10 years)	None	N/A	Egypt
A 5-Year Interval Evaluation of Function after Pollicization for Congenital Thumb Aplasia Using Multiple Outcome Measures [3]	Not Reported	23 months (10 months to 4 years, 9 months)	None	N/A	USA
Children with Surgically Corrected Hand Deformities and Upper Limb Deficiencies: Self-Concept and Psychological Well-Being [4]	Male: 53 Female: 39	10.6 years (9–11 years)	Piers–Harris Children's Self-Concept Scale	Behavior, intellectual and school status, physical appearance and attributes, anxiety, popularity, happiness, and satisfaction	Sweden
Duplicate Thumbs: A Survey of Results in Twenty Patients [5]	Not recorded	3.75 years (1–17 years)	None	N/A	United Kingdom
Low Impact of Congenital Hand Differences on Health-Related Quality of Life [7]	Male: 62 Female: 54	11.8 years (10–14 years)	PedsQL, PUFI	Physical HealthEmotional Functioning Social Functioning School Functioning Manual Ability	Netherlands
Psychological Aspects of Toe to Hand Transfer in Children [12]	Male: 20 Female: 17	8.5 years (2.5–14 years)	None	N/A	United Kingdom
Toe to Hand Transfer in Children: Ten Year Follow Up of Psychological Aspects [11]	Male: 13 Female: 12	Child: 13.29 years (11.95–14.95 years) Teen: 16.92 years (15.75–17.87 years) Adult: 26.60 years (18.03–58.72 years)	CEQ, Hospital Anxiety and Depression Scale, Rosenberg Self Esteem Inventory, Birleson Depression Scale, State Trait Anxiety Inventory, Self- Image Profile,	Depression, anxiety, self-esteem, positive and negative self-image, positive and negative social experiences	United Kingdom
The Psychological Impact of Microvascular Free Toe Transfer for Children and Their Parents [14]	Male: 5 Female: 9	5.08 years (6 months to 13.17 years)	CEQ, Indicators of Parental Adjustment to Congenital Disfigurement, The Perceived Competence Scale, The General Health Questionnaire, The Child Behaviour Checklist,	Positive and negative social experiences, behavioral responses to the baby by the parents, child's subjective sense of functional competence in comparison with his or her peers, maternal anxiety and depression, quantity and quality of children's friendships, their social competence and any behavioral problems	United Kingdom
Hand Function and Activity Performance of Children with Longitudinal Radial Deficiency [15]	Male: 16 Female: 4	Not reported	PUFI	Manual ability	Netherlands

Table 1. continued

Title	Gender distribution	Average age at surgery (range)	Validated survey(s) used	Construct(s) measured	Country
Outcome of Index Finger Pollicisation for the Congenital Absent or Severely Hypoplastic Thumb [16]	Male: 10 Female: 7	12 months (5–54 months)	None	N/A	India
Pollicisation of the Index Finger: A 27 Year Follow Up Study [19]	Male: 6 Female: 5	7 years (2–20 years)	None	N/A	United Kingdom
Outcome after Pollicization: Comparison of Patients with Mild and Severe Longitudinal Radial Deficiency [21]	Male: 16 Female: 8	3.6 years (1.5–15 years)	None	N/A	Netherlands
Comparison of Functional Outcome Scores in Radial Polydactyly [22]	Not reported	1.9 years (less than 1 year to 7 years)	Modified PUFI, ABILHAND-Kids Questionnaire	Manual ability	Netherlands
Perception of the Esthetic and Functional Outcomes of Duplicate Thumb Reconstruction [23]	Male: 17 Female: 14	20 months (range not reported)	None	N/A	USA
Hand Function in Adults with Radial Longitudinal Deficiency [26]	Male: 12 Female: 8	Not reported	QuickDASH, Medical Outcomes Study 12-Item Short-Form Health Survey	Physical Functioning Role Limitations Due to Physical Problems Bodily Pain General Health Perceptions Vitality Social Functioning Role Limitations Due to Emotional Problems Mental Health Upper Extremity Function and Disability	Sweden
Radial Polydactyly: An Outcome Study [31]	Male: 9 Female: 12	7.8 years (1.2–15.8 years)	None	N/A	USA
Objective Features and Aesthetic Outcome of Pollicized Digits Compared With Normal Thumbs [34]	Male: 11 Female: 15	Not reported	None	N/A	USA
Thumb Size and Appearance Following Reconstruction of Radial Polydactyly [37]	Male: 11 Female: 15	Not reported	None	N/A	USA
Central Ray Deficiency: Subjective and Objective Outcome of Cleft Reconstruction [33]	Not reported	Not reported	None	N/A	USA
Complex Syndactyly: Aesthetic and Objective Outcomes [38]	Not reported	Not reported	None	N/A	USA
Functional Outcome After Centralization for Radius Dysplasia [35]	Not reported	22 months (range not reported)	DASH	Upper extremity function and disability	USA

Table 1. continued

Title	Gender distribution	Average age at surgery (range)	Validated survey(s) used	Construct(s) measured	Country
Macrodactyly–Options and Outcomes [41]	Male: 12 Female: 8	62 months	DASH	Upper extremity function and disability	United Kingdom
Long-term Functional Outcome of Patients with Longitudinal Radial Deficiency: Cross-Sectional Evaluation of Function, Activity and Participation [43]	Male: 9 Female: 8	Not reported	Impact on Participation and Autonomy Questionnaire	Autonomy indoors, autonomy outdoors, family role, social relationships, work and educational opportunities	Netherlands
Outcome Measures of Microsurgical Toe Transfers for Reconstruction of Congenital and Traumatic Hand Anomalies [44]	Male: 7 Female: 8	4.9 years (2.1–15.6 years)	Pediatric Outcomes Data Collection Instrument	Upper extremity function, transfer and basic mobility, sports and physical function, pain, comfort, and happiness with physical condition and global function	USA
Toe to Hand Transfer in Children Part 2: Functional and Psychological Aspects [45]	Not reported	Average age not reported (9 months to 14 years)	None	N/A	United Kingdom
Pre-axial Polydactyly: Outcome of the Surgical Treatment [46]	Male: 14 Female: 8	Average age not reported (6 months to 5 years)	None	N/A	Belgium
Comparison of Surgical Treatment and Nonoperative Management for Radial Longitudinal Deficiency [48]	Male: 179 Female: 60	1.5 years	PUFI	Manual ability	India
Long-Term Follow-Up of Surgical Treatment for Thumb Duplication [50]	Male: 10 Female: 8	3 years (10 months to 7 years)	None	N/A	Netherlands
Long-Term Outcomes of Web Creep, Scar Quality, and Function After Simple Syndactyly Surgical Treatment [52]	Male: 18 Female: 1	4.4 years (7 months to 15 years)	None	N/A	Austria
The Preliminary Outcome of Syndactyly Management in Children with a New External Separation Device [56]	Male: 5 Female: 7	Average age not reported (2.6–4.5 years)	None	N/A	China
Functional Outcomes of Children With Index Pollicizations for Thumb Deficiency [58]	Not reported	Not reported	None	N/A	USA
Long-term Results of Surgical Treatment of Thumb Polydactyly [62]	Male: 70 Female: 35	12 months (1–96 months; after exclusion of 1 adult)	None	N/A	Japan
Factors Affecting Surgical Results of Wassel Type IV Thumb Duplications [64]	Male: 29 Female: 12	16 months (6–96 months)	None	N/A	Australia
Functional Assessment After Pollicisation [66]	Not reported	6 years	None	N/A	United Kingdom

Table 1. continued

Title	Gender distribution	Average age at surgery (range)	Validated survey(s) used	Construct(s) measured	Country
Functional Outcome for Children with Thumb Aplasia Undergoing Pollicization [71]	Not reported	2.3 years (1–7.5 years)	None	N/A	USA
Long-Term Outcomes Following Radial Polydactyly Reconstruction [72]	Male: 13 Female: 28	1.1 years (0.4–2.7 years)	DASH, PedsQL	Upper Extremity Function and Disability Physical health Emotional functioning Social functioning School functioning	USA
Comparative Study of Outcomes between Pollicization and Microsurgical Second Toe- Metatarsal Bone Transfer for Congenital Radial Deficiency with Hypoplastic Thumb [73]	Male: 12 Female: 18	3.2 years (18 months to 6 years)	None	N/A	China
The Results of Pollicization for Congenital Thumb Hypoplasia [76]	Male: 21 Female: 14	2.8 years (10 months to 10 years)	None	N/A	Australia
Outcome Analysis, Including Patient and Parental Satisfaction, Regarding Nonvascularized Free Toe Phalanx Transfer in Congenital Hand Deformities [77]	Not reported	4.8 years (6 months to 22 years)	None	N/A	Germany
Index Finger Pollicization in the Treatment of Congenitally Deficient Thumb [78]	Male: 12 Female: 6	3.5 years (1–8 years)	None	N/A	Greece
Thumb Polydactyly: Clinical Outcome After Reconstruction [80]	Male: 24 Female: 10	2.8 years (0.6–47 years)	None	N/A	China
Developing a Pollicization Outcomes Measure [82]	Male: 18 Female: 17	1.9 years (0.5–6 years)	None	N/A	USA

CHD = congenital hand difference; DASH = Disabilities of the Arm, Shoulder, and Hand; PUFI = Prosthetic Upper Extremity Functional Index; CEQ = Childhood Experience Questionnaire; PedsQL = Pediatric Quality of Life Inventory; N/A = not applicable.

hand function (eg, Jebsen-Taylor, dexterity), sensibility, contour, and radiographic appearance (Table 2; Appendix 1 [Supplemental materials are available with the online version of CORR®]).

Results

The PROMs identified in this study covered a mean of 1.3 WHO-ICF domains (SD ± 1.3). These domains include body functions and structures, activity, participation, and environmental factors. Body functions and structures were assessed by 12 of 19 validated measures, whereas 11 captured activity, 11 captured participation, and only six captured environmental factors. Only the Child Behavior Checklist [14] and Piers-Harris Children's Self-Concept Scale [4] captures all four ICF domains. Nine PROMs evaluated only one ICF domain.

Of the 19 validated PROMs used in the 42 studies, only the Prosthetic Upper Extremity Functional Index (PUFI) has been validated specifically in the CHD population (children with congenital longitudinal and transverse deficiency) [7, 8, 22, 48]. Only the ABILHAND-Kids Questionnaire, Disabilities of the Arm, Shoulder and Hand (DASH), Pediatric Outcomes Data Collection Instrument, PUFI, and QuickDASH were developed specifically for use in patients with orthopaedic/upper extremity pathology [7, 8, 22, 26, 35, 44, 48, 72]. The remaining PROMs were developed and validated as generic adult or pediatric outcomes measures. Validated PROMs were used to assess radial longitudinal deficiency (n = 5), microvascular free toe-to-hand transfer (n = 3), all congenital anomalies (n = 2), thumb duplication (n = 2), and macrodactyly (n = 1) (Table 2). The PUFI was used in four studies, the DASH was used in three studies, and the Childhood Experience Questionnaire and Pediatric Quality of Life Inventory were both used in two studies. The remaining instruments were used once.

Patient-reported outcomes measures were used in 13 of the 42 studies. Seven of these 13 studies used more than one instrument and a single study used six different PROMs. Parent-reported outcomes measures were used in six of the 42 studies. Both patient- and parent-reported outcomes were reported in five of the 42 studies.

Discussion

Congenital hand differences profoundly affect a child's physical and psychological development. Although prior research has largely focused on outcomes using objective, functional measures, PROMs capture and quantify the experience of the patient and parents. Ideally, PROMs used to study treatments for CHD would assess psychosocial aspects of the illness as well as symptom intensity and magnitude of disability. In other words, the PROM would address all the WHO-ICF domains. Our purpose was to determine the number of WHO-ICF domains covered by PROMs used in the study of treatments for CHD, the proportion of studies that use PROMs specifically validated among children with CHD, and the proportion of PROMs that target parents, patients, or both.

Our study has several limitations that should be noted. First, CHDs are relatively uncommon, and few studies specifically use PROMs when reporting outcomes for these rare conditions. Additionally, the quality of our findings relies on the quality of the studies included, and these studies vary in the way in which instruments were administered and the study samples that were included. Therefore, we cannot aggregate our data to examine summary effects. Our search only included published articles; other sources such as conference proceedings or unpublished data are not reflected in this review. In this way, there may be newer instruments under development that remain heretofore unexplored but may subsequently emerge to be important measures for future study.

Our review identified that current PROMs used to evaluate patients after surgery for CHD do not fully capture important disability domains as defined by the WHO-ICF framework. This finding likely results from a limited recognition of the impact of psychosocial function on overall health and well-being among those who create survey instruments relevant for children with CHD. Although this four-part model may only provide a broad framework for conceptualizing disability, it likely offers a better overall assessment of how a child with a CHD functions in daily life as compared with alternative measures that focus only on body function/structure or activities. Psychosocial function is highly relevant for children with CHD [42, 81]. In fact, Franzblau et al. [30] found that 58% of children and 40% of parents reported stress related to their CHD. The authors found that 58% of this stress resulted from social interactions, 46% resulted from emotional reactions, and 27% resulted from hand appearance. These factors may be more reliably assessed using a measure designed using the WHO-ICF framework. To this end, disability measures can provide a better assessment of the true outcomes of surgery. Therefore, consideration for each component is important to understand how children and their families experience life with CHD and what elements of disability can and should be addressed with surgery.

Most PROMs we found are not validated for children with CHD. To address the lack of disease-specific items of many of the generic PROMs outcomes instruments, surgeon-generated questionnaires are frequently used.

Table 2. Summary of validated PROMs administered to patients or parents

Name of survey	Number of times used	Construct(s) measured	ICF domain(s)	Number of items	Validated for CHD
Prosthetic Upper Extremity Functional Index	4	Manual ability	Activity	38	Yes
Disabilities of the Arm, Shoulder, and Hand questionnaire	3	Upper extremity function and disability	Body functions and structures, activity, participation	30	No
Childhood Experience Questionnaire	2	Positive and negative social experiences	Participation, environmental factors	20	No
Pediatric Quality of Life Inventory	2	Physical health, emotional functioning, social functioning, school functioning	Activity, participation, environmental factors	23	No
QuickDASH	1	Upper extremity function and disability	Body functions and structures, activity, participation	11	No
Piers-Harris Children's Self-Concept Scale	1	Behavior, intellectual and school status, physical appearance and attributes, anxiety, popularity, happiness, satisfaction	Body functions and structures, activity, participation, environmental factors	80	No
Hospital Anxiety and Depression Scale	1	Depression and anxiety	Body functions and structures	14	No
Rosenberg Self Esteem Inventory	1	Self-esteem	Body functions and structures	10	No
Birleson Depression Scale	1	Depression	Body functions and structures	18	No
State Trait Anxiety Inventory	1	Anxiety	Body functions and structures	20	No
Self-Image Profile	1	Positive and negative self-image, sense of difference, self-esteem	Body functions and structures	25	No
Indicators of Parental Adjustment to Congenital Disfigurement	1	Behavioral responses to the baby by the parents	Participation, environmental factors	6	No
The Perceived Competence Scale	1	Child's subjective sense of functional competence in comparison with his or her peers	Activity, participation	20	No
General Health Questionnaire	1	Maternal anxiety and depression	Body functions and structures	28	No
The Child Behavior Checklist	1	Quantity and quality of children's friendships, social competence, behavioral problems	Body functions and structures, activity, participation, environmental factors	113	No
ABILHAND-Kids Questionnaire	1	Manual ability	Activity	74	No
Impact on Participation and Autonomy Questionnaire	1	Autonomy indoors, autonomy outdoors, family role, social relationships, work and educational opportunities	Activity, participation, environmental factors	31	No
Pediatric Outcomes Data Collection Instrument	1	Upper extremity function, transfer and basic mobility, sports and physical function, pain, comfort, happiness with physical condition and global function	Body functions and structures, activity	86	No
Medical Outcomes Study 12-Item Short-Form Health Survey	1	Physical functioning, role limitations due to physical problems, bodily pain, general health perceptions, vitality, social functioning, role limitations due to emotional problems, mental health	Body functions and structures, activity, participation	12	No

PROMs = patient- and parent-reported outcome measures; ICF = International Classification of Functioning, Disability and Health; CHD = congenital hand difference.

Although these measures provide greater detail regarding outcomes specific to CHD such as web space creep, scarring, and difficulty with specific tasks (eg, buttoning shirts, using scissors), the use of surgeon-generated ad hoc instruments can be problematic. Their frequent use is understandable given that high-quality outcomes measures are extremely complex and require statistical expertise and clinical experience for both creation and application [55]. Surgeon-generated questionnaires are often developed to answer a specific clinical question in the setting of scant resources with a limited emphasis on proper development or validation [10]. If one is to report survey instrument findings, ensuring that the instrument has been validated in the population of interest is an important component of confident data reporting. Furthermore, the ad hoc nature of these instruments generally limits their ability to be used more broadly across other samples and centers for comparability [25]. For example, in our review, only four instruments have been used more than once. As such, it remains difficult to synthesize data across studies using different instruments given the lack of standardized outcomes.

In our review, the majority of PROMs focused on parent-, not patient-reported outcomes for several possible reasons. First, proposed age guidelines for PROMs depend on instrument complexity and the availability of trained assistants for children who may have difficulty reading the material or interpreting a question [20]. In children who are cognitively impaired or otherwise unable to understand survey items, validated parent-proxy reports are available, and many studies record only parent-reported outcomes data. However, discrepancies between child- and parent-reported outcomes have been noted in previous studies with parents either overestimating or underestimating the impact of CHD on their children [2, 3, 8, 10, 40, 44, 55]. One possible way to evaluate the reliability of a proxy report is to administer the exact same PROM in a parallel fashion to both the parental proxy and the child. The correlation between survey responses can be assessed by examining the response means, SDs, and an intraclass correlation coefficient (> 0.7 is considered to be a marker of answer reliability). Second, determining the most appropriate metrics for this unique patient population remains challenging. For example, many outcomes instruments are designed to measure recovery from an injury or some other alteration from a "nondiseased" physical state. Children with CHD have no "normal" baseline with which to compare their current physical and psychological condition. This limitation is further compounded by the substantial adaptability among pediatric patients [4]. In other words, the degree to which a child is affected by a CHD may evolve over time as the child learns to adapt and cope. As such, existing instruments may inadequately measure clinically meaningful differences in long-term outcomes. Lastly, control groups are rarely included in these studies and comparisons with normative data collected during instrument development are typically used. No instruments are available that have been validated in children younger than 5 years of age.

An instrument that captures all the important aspects of biopsychosocial functioning for children with CHD and their families and that is feasible and efficient to administer remains elusive. An ideal PROM for children with CHD would include disease- and age-relevant questions across the full scope of the four WHO-ICF domains. These questions can be based on the comprehensive and brief ICF Core Sets for Hand Conditions developed in 2012 by the multidisciplinary WHO International Consensus Group [67]. The relevance of the survey questions in these Core Sets has been demonstrated [49] and the use of these items allows for a direct content comparison to other measures [28]. Using these categories (eg, emotional functions [body functions], structure of upper extremity [body structures], fine hand use [activities and participation], support and relationships [environmental factors]), a survey may be developed that comprehensively examines the health and well-being of children with CHD. Finally, future studies could explore the need to validate and refine parent-proxy versions that could be used for children who are unable to complete self-reported measures as a result of age or cognitive difficulties.

In conclusion, our systematic review revealed broad heterogeneity in the available PROMs used to capture outcomes among children who have undergone surgery for CHD. Given the profound psychological and sociological sequelae of CHD, a PROM that comprehensively evaluates all biopsychosocial components of disability and is validated for use among children with CHD will improve our ability to measure the effectiveness of treatment and quality of care.

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