

Outcome Tools in Pediatric Foot and Ankle Patients: Comparing Child and Parent Scores

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The study protocol was approved by the Medical Ethics Committee of Seattle Children's Hospital Institutional Review Board (IRB) in Seattle, Washington. Only those families that provided consent to use their responses for research purposes were included in this study, and a separate IRB approval was obtained for this project. The authors certify that they have no affiliations with or financial involvement in any organization or entity with a direct financial interest in the subject matter or materials discussed in this article.

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Abstract

Introduction: The Oxford Ankle Foot Questionnaire for Children (OxAFQ-C) is validated for assessing the impact of foot and ankle conditions in pediatric patients. The purpose of this cross-sectional study is to assess child-parent concordance and identify patient factors that predict improved agreement.

Methods: Patients aged 8 to 16 years with foot and ankle conditions and their parents completed the OxAFQ-C during routine clinic visits over a 9-month period. Demographic and medical information was collected by chart reviews. Responses in each domain were compared using a Wilcoxon signed-rank test, and the comparisons of responses by sex were analyzed with Wilcoxon rank-sum tests. Concordance was assessed with intraclass correlation coefficients.

Results: There were 87 child-parent dyads with 50 female patients (57.5%) and 37 male patients (42.5%). Most parent responders were mothers (84%). The mean patient age was 12.4 (± 2.2) years. The most common diagnosis was pes planus (17%). Child scores were significantly higher than their parents' in the school and play ($P = 0.008$) and emotional ($P = 0.001$) domains. When stratified by age, children younger than 13 years had significantly higher scores than their parents across all domains ($P = 0.015$ physical, 0.002 school and play, 0.001 emotional), although the concordance for the school and play and emotional domains was only moderate (0.73 and 0.58, respectively). Female patients and their parents reported significantly lower scores compared with their male counterparts only in the emotional domain (84.37 vs 93.75, $P = 0.025$).

Conclusion: Concordance is good between child and parent scores of the OxAFQ-C for assessing the impact of foot and ankle conditions. When stratified by age, patients younger than 13 years of age had higher scores than their parents' in all domains with the lowest concordance for the school and play and emotional domains. Female patients and their parents reported significantly lower scores than their male counterparts in the emotional domain.

Level of Evidence: Level IV

Pediatric foot and ankle conditions can lead to notable morbidity. Surgical treatment is typically indicated when there are persistent deformity, consistent pain, and/or functional disability. Determining if a child is experiencing pain and disability can be challenging, and

surgeons must often rely on parents' interpretations of their child's physical and emotional impairment.¹

Over the past 20 years, there is increased interest in health-related quality of life outcomes in the pediatric population.^{2,3} Multiple studies comparing child and parent score agreement have been performed for a variety of medical disorders. The results of these studies range vastly in their outcomes from poor to high child-parent agreement, depending on the disease and outcome being measured. Several studies reported a tendency of children scoring themselves higher than their parents.^{4,5} These patient-reported outcome tools have improved provider ability to evaluate a child's perceived disability. There are little data on the reliability of patient-reported outcomes in children and whether child or parent responses are more accurate.

The Oxford Ankle Foot Questionnaire for Children (OxAFQ-C) is presently the only tool validated for use in pediatric populations to measure the subjective well-being of children aged 5 to 16 years with foot and ankle conditions.⁶ The parental version of the questionnaire is valid for guardians of children aged 5 to 16 years, whereas the child version is administered only to patients aged 8 years or older.⁷ Few published studies compared child and parent-proxy responses for the OxAFQ-C, and they reported only on the concordance regarding specific conditions, such as flexible flat feet and calcaneal apophysitis.⁸⁻¹⁰ Differences among respondents based on factors such as sex and age on parent-proxy child responses for the OxAFQ-C have not been investigated. This information could help providers accurately incorporate parental perceptions in the evaluation of pediatric patients with foot and ankle conditions and improve patient and family expectations.

The purpose of this study is to examine the concordance of self-reported child and parent scores and to identify factors predictive of improved agreement in responses. We hypothesize that the reported scores and concordance will differ based on the age of the patient. We suspect younger children have more difficulty in understanding and communicating to their parents about how their foot and ankle condition affects them, lowering the concordance of their responses.

Methods

Data Collection

From April 2017 until December 2017, the parent and child versions of the OxAFQ-C were administered to the guardians of and the patients aged 8 to 16 years with foot and ankle conditions, respectively, as standard of care to supplement clinical assessment in a single surgeon's tertiary care pediatric orthopedic clinic. An Institutional Review Board-approved prospective database was established, and the parents could consent to their and their children's responses being included for research purposes. Questionnaires were administered to all new patients with foot and ankle problems and their parents/guardians presenting to the senior author's or their physician assistant's clinics. Returning patients and parents repeated the questionnaire at 3, 6, 12 months and then annually after operative or nonoperative intervention. To specifically compare the concordance of child-parent responses, a second Institutional Review Board approval was obtained.

Inclusion criteria for this study were completion of the OxAFQ-C questionnaire by both parent/guardian and child aged 8 to 16 years with a foot and ankle condition. If a child-parent dyad had completed more than one questionnaire during their care,

only the responses of the first survey were included. Of the 93 eligible child-parent pairs, 87 parents (94%) consented to completely participate in this research. All consenting, eligible pairs were included, regardless of underlying diagnosis.

Electronic medical records were reviewed for demographic and medical information including sex, race, ethnicity, International Classification of Diseases (ICD) 9 and 10, and current procedural terminology (CPT) codes.

The Oxford Ankle Foot Questionnaire for Children

OxAFQ-C has been validated to examine the impact of foot and ankle conditions in children.¹¹ The questionnaire consists of 15 5-point Likert scale items, 14 of which are used to assess three domains: physical (6 items), school and play (4 items), and emotional (4 items). Response values and their points value include never (4), rarely (3), sometimes (2), very often (1), and always (0). Domain scores are calculated by deriving the sum of each domain and then dividing by the domain's maximum value. Better function corresponds to a higher score.⁷

Power Analysis

To detect a mean difference between parent and child scores of 5 points with 80% power using a 5%-level 2-sided test, we needed 75 pairs of cases. The current sample contains 87 cases, allowing the detection of potentially even smaller differences (in absolute value).

Statistical Analysis

Normally distributed domain scores were tested using histograms and Shapiro-Wilk tests. The median and interquartile range were calculated for each domain. The parent and child scores were compared using Wilcoxon signed-rank tests. Differences between

female and male patients and parents and the overall comparison between younger (<13 year old) and older (≥ 13 year old) patients were analyzed using Wilcoxon rank-sum tests. Agreement between patient and parent questionnaires was also assessed using intraclass correlation coefficients (ICCs) with a two-way random effects model. As described by Portney et al,^{12,13} ICC reliability was interpreted as <0.5 : poor concordance; 0.5 to 0.75: moderate concordance; 0.75 to 0.9: good concordance; and >0.9 : excellent concordance. This analysis was conducted for the total sample and for the two age groups. Alpha was set at 0.05 to define significance. Statistical analysis was performed using STATA 14.

Results

Eighty-seven pairs of child-parent questionnaires were completed. There were 50 female patient respondents (57.5%) and 37 male patients (42.5%). Of the legal guardians, 84% were mothers, 14% were fathers, and 2% were identified as “other” legal guardians. The mean patient age was 12.4 years (SD 2.2) (Table 1). Diagnosis was recorded using ICD-10 codes with the most common being pes planus in 15 of the 87 children (17.2%) (Table 2).

Only the scores for the physical domain were normally distributed. For consistency, nonparametric Wilcoxon tests are reported throughout. Child scores were higher than parent scores in every OxAFQ-C domain, but they differed significantly only for the school and play ($P = 0.008$) and emotional domains ($P = 0.001$) (Table 2 and Figure 1). ICCs were calculated for parent-child dyads for each domain. Agreement between parent and child scores was rated good in all three domains (physical [0.85], school and play [0.77], and

Table 1

Cohort Demographics

Characteristics	Values
Age (yr)	12.4 \pm 2.4 (8-16)
Females	50 (57.5%)
Males	37 (42.5%)
Completed by	
Mother	73 (84%)
Father	12 (14%)
Unknown/legal guardian	2 (2%)
Pairs	
Mother-son	32 (37%)
Mother-daughter	41 (47%)
Father-daughter	8 (9%)
Father-son	4 (5%)
Unknown-daughter	1 (1%)
Unknown-son	1 (1%)

emotional domains [0.78]), but the concordance was significantly lower for school and play and emotional domains (Table 3).

When stratified by age, children younger than 13 years scored themselves significantly higher than their parents in all three domains (Table 3). There was no significant difference in scores between parents and children aged 13 years and older (Table 3). Similarly, the concordance of scores between younger children and parents (0.84, 0.73, 0.58) was lower than that between patients aged 13 years and older and parents (0.85, 0.81, 0.87) for physical, school and play, and emotional domains, respectively (Table 4).

Female patients reported lower scores in all domains compared with their male counterparts, but only the difference in the emotional domain was statistically significant (84.37 versus 93.75, $P = 0.025$) (Table 5). Parents of female patients also scored their children lower in all domains than parents of male patients, with only the emotional domain differences showing statistical significance (75 versus 93.75, $P = 0.001$).

Table 2

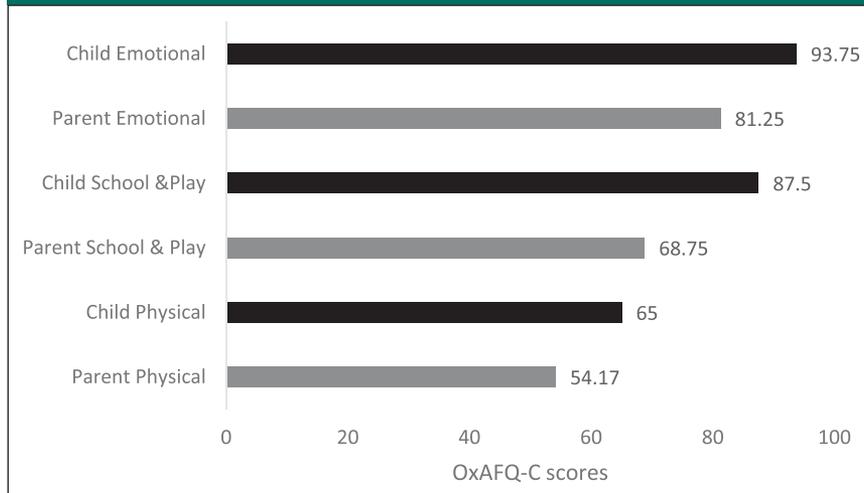
Top 10 Diagnoses

Diagnosis by ICD-10	Values
Pes planus	15 (17%)
Tarsal coalition	9 (10%)
Congenital pes cavus	8 (9%)
Hallux valgus	5 (6%)
Congenital foot deformity	5 (6%)
Idiopathic toe walking	5 (6%)
Fractures of ankle or foot	4 (5%)
Osteochondral lesion	3 (3%)
Freiberg infraction	2 (2%)
Ankle sprain	2 (2%)

Discussion

The OxAFQ-C is the only validated questionnaire for assessing foot and ankle conditions in the pediatric population.¹⁴ Studies of the OxAFQ-C have previously shown good agreement of child-parent responses in multiple languages, validating its use as a health-related quality of life score.^{6,15,16} These studies have not examined whether patient factors might affect concordance. Confirming child-parent concordance is

Figure 1



Median differences between parent and child questionnaires (n = 87).

Table 3

Median, Interquartile Range, and Intraclass Correlation Coefficient for Child and Parent Dyads (All Ages)

	Parent	Child	P Value	Intraclass Correlation Coefficient (95% CI)
Physical domain	54.17 (41.67)	65 (41.66)	0.053	0.85 (0.16-0.90)
School and play domain	68.75 (56.25)	87.5 (56.25)	0.008 ^a	0.77 (0.65-0.86)
Emotional domain	81.25 (31.25)	93.75 (25)	0.001 ^a	0.78 (0.65-0.86)

CI = confidence interval

^a Denotes a statistically significant difference.

important for accurate interpretation of the results. Other pediatric health-related quality-of-life tools have poor child-parent concordance because of a ceiling or floor effect that is common in children.¹⁷ This ceiling effect occurs when children consistently rate their quality of life at the maximal score, regardless of their pathology, or if parents perceive their child experiences disability or suboptimal health status.¹⁸

Our data are consistent with previous validation studies of the OxAFQ-C. There was good agreement between child and parent when evaluating the entire cohort. The concordance was

the highest in the physical domain at 0.85 compared with 0.77 and 0.78 for school and play and emotional domains, respectively. This is most likely because the subject of the physical domain questions is more objective and tangible measure than emotional assessment or ability for play. This is consistent with previous health-related quality-of-life literature that suggests the clinical relevance, such as physical function, of the domain to a specific disease group may be more relevant than the domain itself.¹⁷ Varni et al reported that physical functioning is most important for children with

rheumatoid disease and agreement in this physical domain was highest.¹⁸

Several articles have found evidence that parents of sick children tend to rate their child’s quality of life lower than the child does himself/herself.¹⁹ Children in our cohort similarly rated their function and ability markedly higher than their parents in the school and play and emotional domains. When stratified by age, children younger than 13 years reported statistically significantly higher scores across all three domains. It is unclear at what magnitude a statistical difference in scores correlates to a significant difference in perceived function or limitations. Previous studies rely on the ICCs to determine concordance over absolute values in scores, and we agree this is likely more representative of the true clinical picture.^{6,7,15}

When we stratified the responses by age, concordance for school and play (ICC 0.73) and emotional (ICC 0.58) domains was only moderate between patients younger than 13 years of age and their parents, whereas concordance remained “good” in all domains for older children. Similarly, child scores were significantly higher than parent scores in all domains when analyzing only younger children younger than 13 years of age ($P = 0.015, 0.002, 0.001$). No significant difference was found in the cohort of children aged 13 years or older ($P = 0.761, 0.449, 0.216$) for the physical, school and play, and emotional domains, respectively. We suspect the higher concordance in older children is because of their improved ability to communicate their pain and dysfunction to their parents. We have not found other studies in the literature which investigate age as a factor affecting concordance between patients and parents. This discordance should encourage surgeons to directly discuss with younger patients when assessing their foot and ankle pathology. Additional research into

Table 4**Median, Interquartile Range, and Intraclass Correlation Coefficient for Child and Parent Dyads Stratified by Age**

	Parent	Child	P Value	Intraclass Correlation Coefficient (95% CI)
Children <13 (n = 43)				
Physical domain	54.17 (41.67)	66.75 (29.17)	0.015 ^a	0.86 (0.7-0.93)
School and play domain	68.75 (43.75)	93.75 (31.25)	0.002 ^a	0.73 (0.45-0.86)
Emotional domain	81.25 (37.5)	93.75 (18.75)	0.001 ^a	0.58 (0.21-0.78)
Children ≥13 (n = 44)				
Physical domain	56.25 (41.66)	56.25 (45.83)	0.761	0.85 (0.71-0.91)
School and play domain	68.75 (59.37)	81.25 (59.37)	0.449	0.81 (0.65-0.89)
Emotional domain	75 (28.12)	87.5 (28.12)	0.216	0.87 (0.76-0.93)

CI = confidence interval

^a Denotes a statistically significant difference.**Table 5****Comparison of Median, Interquartile Range (IQR), and Intraclass Correlation Coefficient for Child and Parent Dyads by Patient Sex**

	Median (IQR)—Child			Median (IQR)—Parent of		
	Female (n = 50)	Male (n = 37)	P Value	Female (n = 50)	Male (n = 37)	P Value
Physical domain	58.33 (41.67)	70.83 (29.16)	0.241	47.91 (45.83)	58.33 (25)	0.302
School and play domain	84.37 (58.33)	93.75 (31.25)	0.128	62.5 (56.25)	81.25 (43.75)	0.104
Emotional domain	84.37 (31.25)	93.75 (12.5)	0.025 ^a	75 (18.75)	93.75 (25)	0.001 ^a

^a Denotes a statistically significant difference.

the minimum age at which a child can accurately respond to functional outcome surveys is needed.

When stratified by sex, female patients statistically reported lower scores in the emotional domain compared with their male counterparts (84.37 versus 93.75, $P = 0.025$). This difference was also seen between the parents of female patients and parents of male patients, with significantly lower emotional domain scores in the former (75 versus 93.75, $P = 0.001$). We are not aware of any other literature describing this phenomenon. Mood disorders such as anxiety and depression are more common in female children and adolescents.¹⁷ Perhaps, the comorbidity of a mental health condition in female patients plays a role in their lower emotional scores and their parents' perceptions

of how their foot and ankle condition affects them. Additional research is needed to elucidate the cause of this finding.

This study has several limitations. Examination of the concordance of child-parent scores over time, as well as before and after intervention, would additionally support the use of this outcome tool for children with foot and ankle conditions. We did not have enough patients with longitudinal data in our cohort to evaluate this. Our patient population included children with many different foot and ankle diagnoses. The OxAFQ-C may be less effective or show poorer concordance in certain diagnoses than others. For example, children with cerebral palsy or other neuromuscular conditions may have disabilities or difficulties with gait for reasons other than their foot shape. In addition,

if the child is nonverbal or developmentally delayed, parents may have a harder time interpreting their child's concerns regarding his/her feet.

Conclusion

We demonstrate good child-parent concordance in all three domains of the OxAFQ-C for assessing the impacts of foot and ankle conditions in the pediatric population. When stratified by age, patients younger than 13 years of age had statistically higher scores than their parents' in all domains with the lowest concordance for school and play and emotional domains. Female patients and parents of female patients reported significantly lower scores than male patients and parents of male patients in the emotional

domain. Additional evaluation is needed to confirm these trends. The results of this study serve to improve surgeons' understanding of parent- and child-reported outcome scores for pediatric foot and ankle conditions and to inform their counseling and expectations accordingly.

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