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The Dutch version of the Oxford Ankle and Foot Questionnaire for Children: Useful for evaluation of pediatric foot problems in groups

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ABSTRACT

Background: The purpose of this study is to develop a Dutch version of the Oxford Ankle and Foot Questionnaire for Children (OxAFQ-c) to allow evaluation of pediatric foot care.
Methods: The OxAFQ-c was translated into Dutch, according to the ISPOR-guidelines. Children with different foot and ankle complaints completed the OxAFQ-c at baseline, after two weeks, and after 4–6 months. Measurement properties were assessed in terms of reliability, responsiveness, and construct validity.
Results: Test–retest reliability showed moderate intraclass correlation coefficients. Bland–Altman plots showed wide limits of agreement. After 4–6 months, the group that experienced improvement also showed improved questionnaire outcomes, indicating responsiveness. Moderate correlation between the OxAFQ-c and the Kidscreen and foot-specific VAS-scores were observed, indicating moderate construct validity.
Conclusions: The Dutch OxAFQ-c showed moderate to good measurement properties. However, because we observed limited sensitivity to changes and wide limits of agreement in individual patients, we think the questionnaire should only be used in groups.

1. Introduction

Health-related quality of life questionnaires and Patient-reported Outcome Measures (PROMs) are of increasing interest in clinical research, allowing the opportunity to assess patients' perspective in clinical care [1]. In contrast to provider-focused assessment methods, such as physical examination and clinician-centered rating scales, PROMs can be used to verify subjective experiences of patients. Currently, a distinction is made between generic PROMs and specific PROMs [1]. Generic PROMs are suitable to measure general aspects of health in a broad population, whereas specific PROMs can be targeted for specific populations, diseases, and body regions [2].

Several region-specific PROMs for foot and ankle problems have been developed [3,4]. Nevertheless, most PROMs for foot and ankle problems are developed and validated for adults [4]. Given the differences between the daily activities of children and adults, these PROMs for adults may be less applicable in children with foot and ankle problems.

In 2008, Morris et al. developed the first questionnaire for children with foot and ankle problems, the Oxford Ankle and Foot Questionnaire for Children (OxAFQ-c) [5–7]. This questionnaire is developed for all foot and ankle problems in children and it measures the effect of foot or ankle problems in three domains: (1) physical, (2) school&play, and (3) emotional. Moreover, an extra question about foot wear is included. The questionnaire contains two versions: the OxAFQ-c for 5–17 year old patients and a parent-proxy version.

Unfortunately, the OxAFQ-c is developed in English and is currently not available in the Dutch language. A Dutch version of the OxAFQ-c will contribute to Dutch pediatric ankle and foot care and will give more insights in the children's and parents' perspective of functioning in daily life. However, translated questionnaires should first be validated, otherwise outcomes could lead to invalid conclusions [8]. A good validation procedure

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consists of the assessment of reliability, validity, and responsiveness, in order to ascertain that the translated questionnaire measures reliable and valid outcomes. In 2015, two validation studies of an Italian and a Danish version of the OxAFQ-c were performed, showing good validity and feasibility [9,10]. In order to obtain a validated Dutch version of the OxAFQ-c, we performed in this study a Dutch translation and validation of the OxAFQ-c.

2. Patients and methods

2.1. Translation

The linguistic translation of the OxAFQ-c contained a forward and backward translation performed according to the ISPOR guidelines for translation [11]. After translation a consensus meeting was planned with the forward translators and principal investigator to establish a first version of the Dutch child and proxy questionnaires. Comprehensibility of these questionnaires was tested at the orthopedic outpatient clinic of the Sophia Children's Hospital. Patients with foot and ankle problems and their parents were asked to complete the translated questionnaire. Comments on difficulties in this translated questionnaire were incorporated in the final version of the Dutch OxAFQ-c.

2.2. Validation process

2.2.1. Patient selection

Prior to the inclusion of patients, the study was approved by the Institutional Review Board of the Erasmus Medical Centre, Rotterdam (MEC-2014-669). Children between the age of 5 and 17 years old with all kinds of foot or ankle complaints and their parents were recruited at the orthopedic outpatient clinic and the orthopedic operation department of the Sophia Children's Hospital, Rotterdam. The included population consisted of patients that were waiting for surgery, were already treated and came for a clinical control appointment, or were newly admitted to the outpatient clinic. Informed consent was signed by parents of children that participated in this study. Children above the age of 12 years old also signed informed consent.

2.2.2. Measurements

All included patients and their parents were asked to complete the OxAFQ-c three times; the first time at the outpatient clinic or before surgery, the second time two weeks after the first questionnaire, and the third time four to six months after the first questionnaire. Two weeks was considered as sufficient time to assume patients could not remember their answers, but also no change in foot complaints occurred. Four to six months was considered as sufficient time to show improvement of the complaints. The second and third questionnaires were sent by mail to all participants. When no response was received, patients were called for a reminder.

The OxAFQ-c is a questionnaire for children with foot and ankle problems and contains 15 questions in four different domains: physical (6 questions), school&play (4 questions), emotional (4 questions), and footwear (1 question) [5]. The questionnaire is available in a child-version and proxy-version. All questions have five answer options, scoring from zero (always) to four (never). Per domain a score can be calculated by dividing the total amount of points by the maximal amount of points in that domain. A high score represents a better outcome [5].

2.2.2.1. Reliability. Reliability is defined by the COSMIN panel as 'the degree to which the measurement is free from measurement error' [8], and was analyzed by test–retest reliability and internal consistency. Test–retest reliability was evaluated by comparing the first and second scoring of the questionnaire, assuming no difference between foot and ankle function between these two moments. Patients who already received treatment at the second measurement were excluded from analysis. As it is suggested that questionnaires in younger children are less reliable [5], we additionally analyzed the effect of age on measurement error by excluding children below the age of eight. Internal consistency was used to evaluate if each question in one domain measured the same construct. To do so, correlation between questions in every domain was studied, testing for sufficiently strong relations between the items to assume that they measure the same construct.

2.2.2.2. Responsiveness. Responsiveness is defined by the COSMIN panel as 'the ability of an instrument to detect change over time in the construct to be measured' [8]. Therefore, all patients that completed the third OxAFQ-c were also asked if foot complaints became worse, better, or stayed the same compared to the first measurement time. Differences in domain outcomes between the first and third OxAFQ-c were compared between the three complaint groups in order to evaluate if the OxAFQ-c was able to detect change over time in those patients that reported a change.

2.2.2.3. Construct validity. Construct validity is defined by the COSMIN panel as 'the degree to which the scores of a measurement instrument are consistent with hypotheses (regarding internal relationships, relationships with scores of other instruments or differences between relevant groups)' [8]. To test construct validity, seven hypotheses were formulated about the relationship between the OxAFQ-c and two additional questionnaires; the Kidscreen and foot-specific VAS-scores. All questionnaires were completed at baseline.

The Kidscreen is a generic health related quality of life questionnaires, which is validated in the Dutch language [12]. Because the developers have used the 27-item questionnaire for the development of the OxAFQ-c to test construct validity, we chose to use this questionnaire in our validation study. All questions have five answer options, scoring from zero (never) to four (always). According to the questionnaire manual, the domain scores were recalculated in a way that higher scores indicate higher quality of life.

Additionally, four different VAS-scores for foot pain, foot function, foot appearance, and influence of foot complaints on daily life were scored by children and parents. These VAS-scores were included to be able to formulate a sufficient amount of hypotheses to test construct validity [8]. In the VAS, zero was equal to absence of pain, perfect function, maximal satisfaction about appearance, and no problems in daily life, ten was equal to maximal pain, no function, no satisfaction about appearance, and always problems in daily life. The seven hypotheses about the relationship between the OxAFQ-c and the additional questionnaires were:

1. A higher score in the physical domain of the OxAFQ-c correlates with a higher score in the physical domain of the Kidscreen;
2. A higher score in the school&play domain of the OxAFQ-c correlates with a higher score in the peers and social support domain of the Kidscreen;
3. A higher score in the emotional domain of the OxAFQ-c correlates with a higher score in the psychological well-being domain of the Kidscreen;
4. A higher score in the physical domain of the OxAFQ-c correlates with a lower VAS-score on foot function;
5. A higher score in the physical domain of the OxAFQ-c correlates with a lower VAS-score on pain;
6. A higher score in the school&play domain of the OxAFQ-c correlates with a lower VAS-score on daily life;
7. A higher score in the emotional domain of the OxAFQ-c correlates with a lower VAS-score on appearance.

Table 1
Patient characteristics.

Characteristics	Baseline (SD)	Second questionnaire (SD)	Third questionnaire (SD)
No. of patients	64	41	36
Gender (M/F)	37/27	22/19	24/12
Age (SD) (in years)	10.8 (3.4)	11.1 (3.0)	11.1 (3.0)
Diagnosis (no. of patients)			
Congenital talipes equinovarus	18	11	14
Idiopathic toe-walker	5	1	2
Pes planus/pes planovalgus	5	4	3
Trauma	8	5	4
Tarsal coalition	6	6	1
Pes cavovarus (due to HMSN, CP, sacral Ewing sarcoma)	7	5	4
Toe deformities (hallux valgus, macrodactyly, M. Freiberg)	6	3	5
Tumor or exostoses (lipoma, tarsal bossing, multiple hereditary exostoses)	3	3	1
Other (osteochondritis dissecans talus, idiopathic pain, pes transversus, pes equinus, cleft foot)	6	3	2
	Mean domain scores at baseline (range)	Mean domain scores 2nd questionnaire (range)	Mean domain scores 3rd questionnaire (range)
OxAFQ — child			
Physical	52.2 (0–100)	43.1 (0–100)	52.3 (0–100)
School & play	76.7 (0–100)	61.3 (0–100)	71.2 (12.5–100)
Emotional	78.5 (31.3–100)	74.1 (18.8–100)	74.7 (6.3–100)
Foot wear	58.6 (0–100)	49.4 (0–100)	52.8 (0–100)
OxAFQ — parent			
Physical	48.0 (0–100)	50.4 (0–100)	55.5 (0–100)
School & play	70.0 (6.3–100)	69.2 (0–100)	75.4 (31.5–100)
Emotional	74.1 (37.5–100)	74.8 (25–100)	73.4 (0–100)
Foot wear	55.1 (0–100)	60.4 (0–100)	59.0 (0–100)

2.3. Statistical analysis

Domain scores of the OxAFQ-c were treated as continuous data. Normal distribution of domain scores and VAS-scores were tested by histograms and Kolmogorov–Smirnov tests. All tests were two-tailed and statistical significance was assumed at $p < 0.05$. A missing data-analysis was performed after collection of all questionnaires. Percentage of missing questions were calculated and Little's tests was used to test for missing completely at random (MCAR). The imputation method was determined based on these outcomes. Statistical analyses were performed using IBM SPSS 22.0.

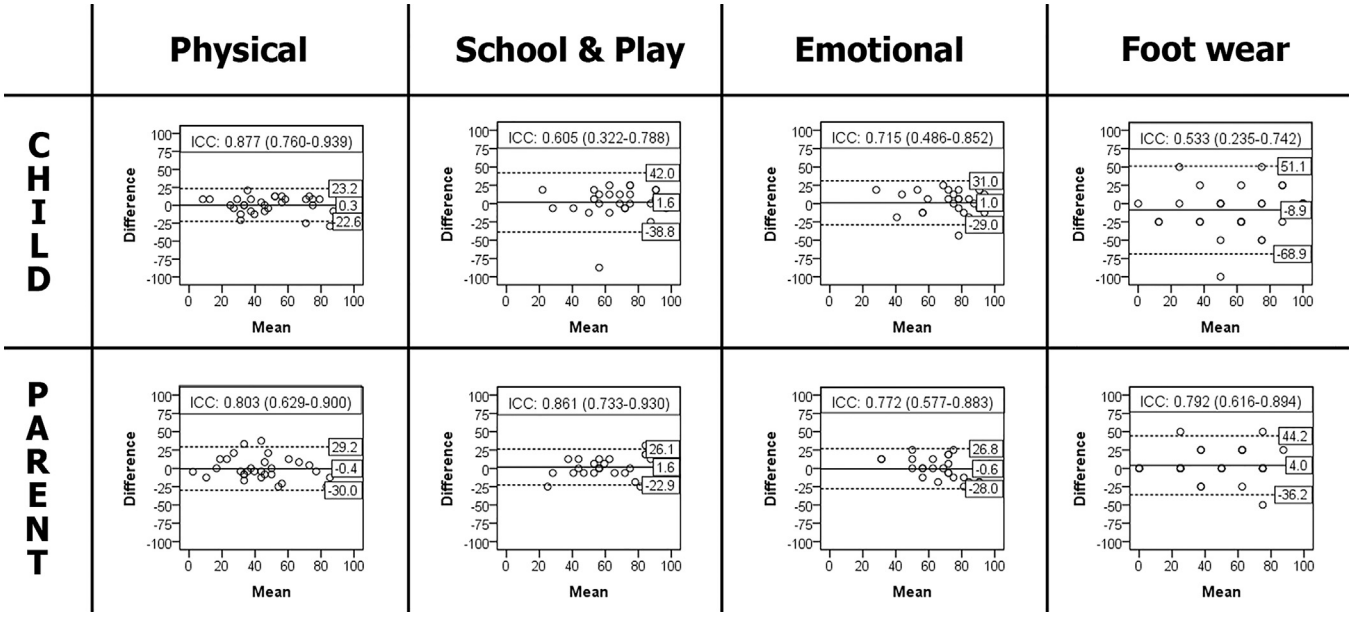

Fig. 1. Bland–Altman plots and ICCs per domain of the parent and child version of the OxAFQ-c in children between the age of 5 and 17 years.
Solid lines represent mean differences; dotted lines represent limits of agreement.

Table 2
Mean differences, limits of agreement and ICCs between the first and second questionnaire in children aged 8 years and older.

Children >7 years (n = 24)		Mean difference (SD difference)	Limits of agreement	ICC (95% CI)
Child	Physical	−0.3 (11.3)	−22.4 to 21.8	0.884 (0.750–0.948)
	School & play	−2.1 (21.5)	−44.2 to 40.0	0.579 (0.233–0.794)
	Emotional	1.8 (15.5)	−28.6 to 32.2	0.714 (0.444–0.865)
	Foot wear	−11.5 (30.4)	−71.1 to 48.1	0.538 (0.194–0.767)
Parent	Physical	−0.02 (16.0)	−31.4 to 31.3	0.764 (0.524–0.891)
	School & play	0.3 (13.5)	−26.2 to 26.8	0.831 (0.647–0.923)
	Emotional	−1.0 (13.0)	−26.5 to 24.5	0.797 (0.585–0.907)
	Foot wear	3.1 (20.0)	−36.1 to 42.3	0.818 (0.626–0.917)

2.3.1. Reliability

Test–retest reliability was analyzed with use of intra-class correlation coefficients (ICC) and Bland–Altman plots per domain of the OxAFQ-c [13]. ICCs for all test–retest analyses were calculated with a two-way random effect model, absolute agreement. ICCs above 0.7 were considered good [13]. Mean scores of the first and second measurement were plotted against the difference between these two measurements to obtain Bland–Altman plots [14]. Systematic and random measurement error were assessed with use of respectively the mean difference and limits of agreement. Limits of agreement were calculated as the mean difference plus or minus 1.96× the standard deviation of the mean difference. Because we expected no changes in foot and ankle complaints between the two measurements, the mean difference should be close to zero combined with a small interval between the limits of agreement [14].

Internal consistency per domain was tested with Cronbach's alfa [15]. Cronbach's alpha's between 0.7 and 0.9 were considered good [13].

2.3.2. Responsiveness

Differences in domain score between the first and third questionnaire were calculated and plotted per complaint group. We expected a difference above zero in the group with improved foot and ankle complaints and no difference of domain score in the group without improvement.

2.3.3. Construct validity

Correlation between the OxAFQ-c, the Kidscreen and VAS-scores was calculated with the Spearman correlation coefficient (r). Values were rated as: $r < 0.3$ = low correlation, $r = 0.3$–0.49 = medium correlation, and $r > 0.50$ = high correlation [5].

3. Results

3.1. Translation

Forward and backward translations did not lead to differences in the most crucial words of the questionnaire. The English questionnaire used different verb tenses than the Dutch questionnaire. Therefore, at the consensus meeting we decided to change the verbs to create a more similar meaning of the questions. At the test phase, seven children and their parents completed the questionnaires. They did not report problems in understanding the questionnaire.

3.2. Validation process

3.2.1. Patient selection

Sixty-four patients and their parents completed the first questionnaire. Twenty-seven girls and 37 boys were included with a mean age of 10.8 (SD = 3.4) and a variety of foot and ankle disorders (Table 1). The second questionnaire was completed by 41 patients and their parents (response rate: 64%). The third questionnaire was completed by 36 patients (response rate: 56%). No differences in age and gender were seen between baseline, the second group (gender: p = 0.676; age: p = 0.555) and the third group (gender: p = 0.595; age: p = 0.384).

3.2.2. Missing data

Fifteen questions of the OxAFQ-c (0.7%) were missing in the completed children questionnaires at all three time points, ten questions (0.5%) were missing in the parent questionnaires. Missing data was equally distributed among the questions. Little's test showed that data were MCAR (p = 0.801). Together with the small proportion of missing data, we decided to perform single imputation of the domain scores using the expectation maximization algorithm. This type of imputation provides unbiased estimates and improves statistical power of analyses [16].

3.2.3. Reliability

The four different domains showed moderate to good ICCs between the first and second questionnaire. However, Bland–Altman plots showed an extensive random variability indicated by vast limits of agreement (Fig. 1). Because ten patients already received treatment at the time of the second questionnaire, 31 patients were included in this reliability analysis. Excluding younger patients did not improve the test–retest reliability (Table 2).

Cronbach's alpha of both parent and child versions of the physical and school&play domains were above 0.7 (Table 3). The emotional domain had an alpha lower than 0.7, caused by the question about walking pattern. Exclusion of this question led to an alpha above 0.7 in both versions.

3.2.4. Responsiveness

Mean differences of the group that experienced improvement of foot and ankle complaints were above zero in all domains (Fig. 2), indicating that domain scores increased between the first and third questionnaire. The group that experienced no change in foot and ankle complaints showed in

Table 3
Internal consistency between questions per OxAFQ-c domain, evaluated with Cronbach's alpha.

Version		Cronbach's alpha
Child	Physical	0.826
	School & play	0.858
	Emotional	0.602
	Foot wear	–
Parent	Physical	0.915
	School & play	0.906
	Emotional	0.671
	Foot wear	–

Values below 0.7 indicate that one or more questions in that domain do not measure the same construct.

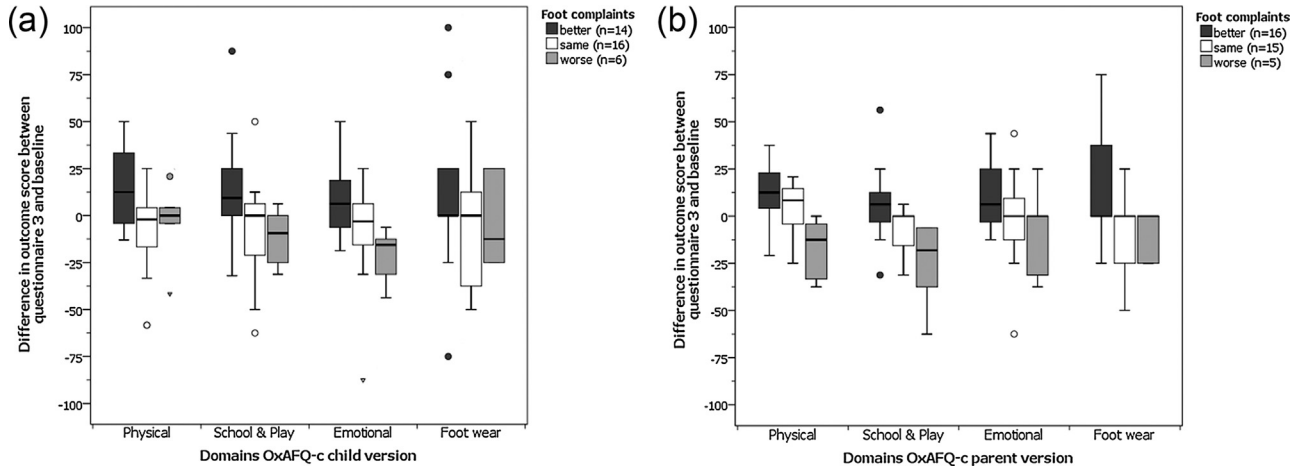

Fig. 2. Mean difference between third questionnaire and baseline questionnaire in different foot complaints groups per domain of the child and parent version of the OxAFQ-c.

all domains a score around zero and was in all domains lower than the improved group.

3.2.5. Construct validity

Six of the seven hypothesis showed medium to high correlations. All lower VAS-scores were correlated with a higher OxAFQ-c domain score (Table 4). The correlation between the school&play domain of the OxAFQ-c and the peers and social support domain of the Kidscreen showed a non-significant correlation.

4. Discussion

To develop a Dutch questionnaire for children with foot and ankle complaints, we translated the English OxAFQ-c into the Dutch language and validated this new questionnaire. To validate the questionnaire, we tested reliability, responsiveness, and construct validity. Test–retest reliability was assessed by comparing the baseline OxAFQ-c with the OxAFQ-c completed after two weeks. Outcomes showed ICC's above 0.7, indicating a good test–retest reliability. When comparing baseline outcomes of the OxAFQ-c with outcomes after 4–6 months, we found a clear difference in outcomes in patients that reported improvement and patients that reported no difference in foot or ankle complaints, indicating responsiveness to change of the questionnaire. Construct validity was assessed by testing seven hypotheses regarding the relationships between the OxAFQ-c and other questionnaires, showing a significant correlation in six of the seven hypotheses. Altogether, measurement properties of the Dutch version of the OxAFQ-c were moderate to good and we therefore concluded that the questionnaire is useful for evaluation of foot and ankle problems in pediatric patient groups.

The Dutch translation showed good test–retest reliability, reflected by ICC's above 0.7 in most domains. However, analysis with Bland–Altman plots showed vast limits of agreement, indicating that domain scores can differ more than 25 points in one patient, without changes in foot and ankle complaints. This suggests that the questionnaire will not be very sensitive or even incapable of evaluating improvement of foot and ankle complaints in individual patients, since changes up to around 25 points may be attributed to change [14]. This finding is not new, although it was

Table 4
Spearman's correlation of the seven pre-formulated hypotheses regarding the relationships between domain scores of the OxAFQ-c and the Kidscreen and VAS-scores.

Version	Hypothesis	Spearman correlation coefficient	p-Value	Hypothesis confirmed
Child	A higher score in the physical domain of the OxAFQ correlates with a higher score in the physical domain of the Kidscreen.	0.578	<0.001	√
	A higher score in the emotional domain of the OxAFQ correlates with a higher score in the psychological well-being domain of the Kidscreen.	0.381	0.002	√
	A higher score in the school and play domain of the OxAFQ correlates with a higher score in the peers and social support domain of the Kidscreen.	−0.025	0.846	
	A higher score in the physical domain of the OxAFQ-c correlates with a lower VAS-score on foot function.	−0.526	<0.001	√
	A higher score in the physical domain of the OxAFQ-c correlates with a lower VAS-score on pain.	−0.596	<0.001	√
	A higher score in the school and play domain of the OxAFQ-c correlates with a lower VAS-score on daily life.	−0.382	<0.001	√
	A higher score in the emotional domain of the OxAFQ-c correlates with a lower VAS-score on appearance.	−0.375	0.002	√
Parent	A higher score in the physical domain of the OxAFQ correlates with a higher score in the physical domain of the Kidscreen.	0.543	<0.001	√
	A higher score in the emotional domain of the OxAFQ correlates with a higher score in the psychological well-being domain of the Kidscreen.	0.390	0.001	√
	A higher score in the school and play domain of the OxAFQ correlates with a higher score in the peers and social support domain of the Kidscreen.	−0.041	0.747	
	A higher score in the physical domain of the OxAFQ-c correlates with a lower VAS-score on foot function.	−0.558	<0.001	√
	A higher score in the physical domain of the OxAFQ-c correlates with a lower VAS-score on pain.	0.667	<0.001	√
	A higher score in the school and play domain of the OxAFQ-c correlates with a lower VAS-score on daily life.	−0.614	0.002	√
	A higher score in the emotional domain of the OxAFQ-c correlates with a lower VAS-score on appearance.	−0.484	<0.001	√

never specifically underlined. For example, the Danish translation of the OxAFQ-c showed comparable results, with limits of agreement as large as −1.69 to 1.99 on a scale of −4 to 4 [9]. Unfortunately, comparison with the results from the original developers is not possible, because they did not report Bland–Altman plots or Smallest Detectable Changes (SDC) [5]. Nevertheless, the good ICCs, absence of systematic errors, and the mean differences around zero in the Dutch questionnaire suggest that the questionnaire is useful for evaluation of foot and ankle complaints in pediatric patient groups. Notably, practical application of the Dutch OxAFQ-c in its current form is limited to the assessment of general function or treatment outcomes at the population level. Its use at the individual patient level is hampered by the wide limits of agreement, which might therefore introduce measurement error. This is likely true of a myriad of patient-reported outcome measures.

Internal consistency showed three alpha's outside the proposed interval of 0.7–0.9. In the emotional domain an alpha below 0.7 was observed, caused by the question about walking pattern. Exclusion of this question resulted in an alpha above 0.7, which might indicate that this question does not measure the same construct as the rest of the questions in this domain. The physical and school&play domain of the parent version had both an alpha slightly above 0.9, indicating that questions in the specific domains are very related and may not have an additional contribution to the domain score. Because other translations studies did not show comparable results in their populations [9,10] and higher alpha's up to 0.95 are also accepted in literature [17], we did not remove these questions.

Evaluation of responsiveness showed that the group with improvement of their foot and ankle complaints, as measured with a single question about improvement, had higher domain scores in the third measurement compared to the group that reported similar foot and ankle complaints. This indicates that the questionnaire is capable of detecting improvement of the foot and ankle complaints, which is in line with the conclusions of the other translations of the OxAFQ-c [9,10]. Nevertheless, differences between the groups were small and should be interpreted with caution since reliability analysis showed that large differences in domain scores in individual patients may occur without changes in complaints.

The Dutch version of the OxAFQ-c is a comprehensible questionnaire, also for young children. Exclusion of children below the age of eight did not result in better reliability. Furthermore, only small differences in domain scores between parents and children were observed, which suggests that children understand the questionnaire. Although the direction and magnitude of the relationship between the domain scores of children and their parents is unknown, Ardon et al. showed in congenital hand differences that mean group scores of parents and children did not differ a lot, which is similar to our results [18].

To reduce the burden to participate in the study, administration of the questionnaires took place in both the hospital and at home, which may have influenced the outcomes. The first questionnaire was often completed in the hospital, where focusing on the foot or ankle problem during the visit could have led to a systematical lower score on the OxAFQ-c. The second and third questionnaire were completed at home, without the additional focus on the foot or ankle problem, leading to a possible higher outcome. Despite this potential effect, mean differences between the first and second questionnaire were two-sided, showing both positive and negative differences in different domains. This indicates no consistent pattern in reporting higher or lower scores at a specific administration moment.

Unfortunately, during the study we experienced a lower response rate than anticipated, which resulted in a smaller sample sizes at the second and third measurement moment than expected. Furthermore, sample size of the second measurement was decreased due to the fact that some patients received treatment within two weeks after the first questionnaire. Nevertheless, the results of the test–retest analysis show that comparing outcomes of individual patients might introduce measurement error. Inclusion of more patients would not have changed the outcomes of the Bland–Altman plots in a way that the questionnaire will be useful for comparing individual patients.

5. Conclusions

We translated and validated the first questionnaire for children with foot and ankle problems into the Dutch language. Comparable with previous studies, analysis of test–retest reliability indicates that the OxAFQ-c is not very sensitive to observe changes in individual patients and therefore should carefully be used in individual children with foot and ankle problems. Nevertheless, we think the OxAFQ-c has sufficient measurement properties to compare outcomes or identify foot and ankle problems in pediatric patient groups, allowing evaluation of patient's perspective in Dutch pediatric foot and ankle care.

Availability of the questionnaire

For more information about the Dutch Oxford Ankle and Foot Questionnaire for children go to: https://innovation.ox.ac.uk/clinical-outcomes/patient-reported-outcome-measures/. For license applications go to: https://process.innovation.ox.ac.uk/.

Conflict of interest

Authors declare that they have no conflict of interest.

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