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Vancouver Symptom Score for Dysfunctional Elimination Syndrome: Reliability and Validity of the Dutch Version

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Purpose: We sought to establish the reliability and validity of the Dutch version of the Vancouver Symptom Score for Dysfunctional Elimination Syndrome for children with dysfunctional voiding and their parents.

Materials and Methods: For this cross-sectional multicenter study the Vancouver Symptom Score for Dysfunctional Elimination Syndrome was translated and cross-culturally adapted to Dutch following a standardized process. Patients 16 years or younger with dysfunctional voiding and their parents were recruited at pediatric, pediatric urology and pelvic floor physical therapy outpatient clinics. The reference group consisted of children 6 to 16 years old without dysfunctional voiding and their parents. All groups completed questionnaires. The evaluated measurement properties included discriminative ability, internal consistency, test-retest reliability, interrater agreement, criterion validity using the Pediatric Incontinence Questionnaire and construct validity. A cutoff value for diagnosis of dysfunctional voiding was determined.

Results: A total of 50 patients and 60 references and their parents were included in the study. The Vancouver Symptom Score for Dysfunctional Elimination Syndrome showed good discriminative ability. A moderate internal consistency was found (Cronbach alpha 0.37-0.55). Test-retest reliability was moderate to good, and interrater agreement demonstrated good correlation between children and parents (ICC 0.85, 95% CI 0.79–0.89). A weak correlation with the Pediatric Incontinence Questionnaire was found in patients and construct validity was confirmed. Cutoff scores for dysfunctional voiding were 11 and 9 for patients and parents, respectively.

Conclusions: The Dutch Vancouver Symptom Score for Dysfunctional Elimination Syndrome displayed moderate to good reliability and validity properties for the patient and parent versions. Use of this instrument in clinical practice will support the assessment of dysfunctional voiding and facilitate international reporting of research results.

Key Words: pediatrics, surveys and questionnaires, urination disorders

Abbreviations and Acronyms

DV = dysfunctional voiding

ICC = intraclass correlation coefficient

PiNQ = Pediatric Incontinence Questionnaire

pPiNQ = Parent Pediatric Incontinence Questionnaire

pVSSDES = Parent Vancouver Symptom Score for Dysfunctional Elimination Syndrome

QoL = quality of life

VSSDES = Vancouver Symptom Score for Dysfunctional Elimination Syndrome

URINARY incontinence is present in about 7% of children at age 7 years.[1] Dysfunctional voiding is the underlying cause in approximately 30% of cases.[2] Symptoms of dysfunctional voiding other than urinary incontinence include recurrent urinary tract infections due to high post-void

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Study received local ethics committee approval (MEC-2014-290).

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residuals.[3] Bowel dysfunction is also strongly associated with dysfunctional voiding as 50% of children with dysfunctional voiding present with constipation or fecal incontinence.[4] Therefore, treatment of dysfunctional voiding is targeted at improving bladder and bowel symptoms simultaneously.

Using questionnaires to assess the severity of symptoms is increasingly recommended.[5] Different scoring systems to classify DV have recently been developed,[6–8] although no questionnaire for this evaluation currently exists in Dutch. A Dutch questionnaire to evaluate the presence of urinary incontinence exists but it does not discriminate for diagnosis.[9]

We chose to validate the VSSDES because it has undergone a stringent validation process in the original development phase and displayed adequate measurement properties.[10] In addition, it is a short questionnaire that also addresses bowel symptoms, and cutoff values have been established, indicating diagnostic ability. Other scoring systems fulfill some but not all of these criteria. The VSSDES has also proved to be effective for assessment of DV in a larger cohort in the United States.[11] Therefore, the VSSDES can function as a tool to support diagnosis of DV and potentially assess effectiveness of treatment. Furthermore, combined use of the VSSDES with the validated Dutch PiNQ,[12] which evaluates the impact of urinary incontinence on quality of life in children, would enable Dutch clinicians to comprehensively assess the severity and impact of DV on quality of life and evaluate the effects of treatment. We translated the VSSDES into Dutch to assess the psychometric measurement properties of the Dutch version, thereby meeting the need for a Dutch standardized measure for this heterogeneous syndrome.

MATERIALS AND METHODS

Study Group

Approval was obtained from the local ethics committee for this prospective cross-sectional study (MEC-2014-290). Patients were asked to participate in the study by their treating physician. Inclusion criteria for children were having DV (based on clinical symptoms, bladder and bowel diaries, uroflowmetry with a staccato and/or intermittent flow, and determination of residual urine), being 16 years or younger and being fluent in Dutch. Also at least 1 parent should be fluent in Dutch. Exclusion criteria were known anatomical abnormalities of the urinary tract, neurogenic disease and previous urological surgery.

All consecutive children with DV and their parents presenting between October 2014 and August 2015 at the outpatient clinics of pediatrics, pediatric urology and pelvic floor physical therapy at 2 community hospitals and 1 tertiary hospital who fulfilled these criteria were asked to participate in the study. Written informed consent was obtained from parents and from patients if they were 12 years or older. Subsequently patients and parents were asked to complete the first of 2 questionnaires. The second questionnaire was completed at home 1 to 2 weeks after inclusion and returned through the postal service.

The reference group consisted of children and their parents attending a local primary school who were randomly approached for study participation. Children 6 to 16 years old without a known urological history or neurogenic disease who were fluent in Dutch (as well as at least 1 of their parents) were eligible for inclusion. After obtaining written informed consent children and parents were asked to fill out the questionnaires.

Questionnaires

The questionnaire contained 2 measures. The VSSDES is a 14-item condition specific measure to evaluate symptoms of bladder and bowel dysfunction. The last item, which evaluates the difficulty of the measure, is not included in the score. All remaining items are weighted equally and all item responses except 1 are scored using a 5-point Likert scale, with scores ranging from 0 (no complaints) to 4 (severe symptoms). Total scores range from 0 to 52, with higher scores indicating more severe symptoms.[10] In the current validation study all patients (VSSDES) and parents (pVSSDES) were asked to complete the questionnaire themselves when possible. The PiNQ is a condition specific measure to evaluate health related QoL in children with urinary incontinence. The measure consists of 20 items and is available in a pediatric (PiNQ) and a parent (pPiNQ) version. Higher scores indicate lower QoL.[12]

Linguistic Validation

The VSSDES was translated and cross-culturally adapted following standardized guidelines.[13] The English version of the VSSDES was independently translated to Dutch by 3 native Dutch speakers. A discussion and revision of differences in translation resulted in a consensus version, which was then translated backward by a native English speaker and compared with the original version. A face-to-face test was performed with 10 children and their parents, and changes were made accordingly, resulting in the final Dutch version (supplementary Appendix, http://jurology.com/).

Measurement Properties and Reliability

The Dutch VSSDES was validated by means of quality criteria for standardized measurement properties.[14] For the reliability analysis Cronbach alpha was calculated to determine internal consistency for the patient and reference group. To assess the reproducibility, the ICC for agreement was calculated to evaluate test-retest reliability in the patient group.[15] In addition, limits of agreement were reported, which equal the mean change in scores of repeat measurement $\pm$ 1.96 SD.[16] Interrater agreement was determined by calculating the ICCs and Pearson correlations between the children and parent scores for both groups.[17]

Validity

For the validity analysis the content validity was evaluated during linguistic validation by patients and

researchers. No real gold standard exists for the clinical symptoms of DV to assess the criterion validity. The PiNQ was used as a substitute since higher scores on the PiNQ have been associated with the presence of DV.[12] Spearman rho was used to calculate the correlation between the VSSDES and the PiNQ. The hypotheses that were predefined to determine the construct validity were 1) patients will have higher scores on the VSSDES compared to the reference group, 2) patients with higher scores on the VSSDES will have higher scores on the PiNQ, and 3) children and parent scores on the VSSDES will correlate with each other. A principal component analysis was performed to examine the 4 dimensions of the VSSDES found in the original validation study.[10]

Statistical Methods

A sample size of at least 50 participants is considered adequate for validation of questionnaires.[14] Therefore, we aimed to include a total of 100 children, ie 50 patients and 50 references. Differences between patient and reference groups, and patient and parent scores were evaluated using the Mann-Whitney U test and chi-square test for continuous and categorical variables, respectively. A ROC curve was created to determine if the optimal threshold score in our patient group was similar to the one in the original validation article, which was 11.[10] A 2-sided p value of less than 0.05 was considered significant. Statistical analyses were performed using IBM SPSS Statistics®, version 21.0.

RESULTS

The study population consisted of 110 participants and their parents. In the patient group 50 children and their parents completed the first set of questionnaires, and 31 patients and parents completed the second set of questionnaires. The reference group included 60 children and their parents. [T1] Table 1 displays the patient and reference characteristics. There was no significant difference in age or gender between patients and references. Median baseline VSSDES scores were similar between patient and reference groups, as were pVSSDES scores, revealing the discriminant ability of this instrument. The VSSDES was classified as very easy or easy by 64% of the children and 69% of the parents. In 1 patient-parent pair the patient was unable to complete the VSSDES independently and only the pVSSDES was available because of the age of the child.

Reliability

The internal consistency was moderate with a Cronbach alpha of 0.55 and 0.55 for patients, and 0.42 and 0.37 for references, for the VSSDES and pVSSDES, respectively. These findings confirm the results of the original validation study. The test-retest reliability in 32 patients and parents was moderate, with ICC values of 0.41 (95% CI 0.07–0.66) and 0.41 (95% CI 0.07–0.67) for the VSSDES and pVSSDES, respectively. The test-retest period (median 15 days, IQR 5–33) in the present study was longer than in the original study (1 week), in which nonstable patients could have been included. Respective ICCs increased to 0.94 (95% CI 0.80–0.98) and 0.79 (95% CI 0.45–0.93) when only responses with a 1-week test-retest period were selected. Interrater agreement between children and parent scores is shown in table 2. [T2] For the total group good correlations were found between child and parent scores for ICC values. When considering the age of 9 years as a cutoff for DV in the patient group, the correlation is strengthened in the group younger than 9 years but there is only a weak correlation between patients and parents in the group older than 9 years.

Validity

Content validity was considered adequate during the linguistic validation process and face-to-face test by patients and researchers. The criterion validity was assessed by evaluating the correlation between the VSSDES and PiNQ. In children a weak correlation ($r = 0.31$, $p = 0.04$) and in parents no correlation ($r = 0.09$, $p = 0.56$) was found. Consequently the second hypothesis could not be confirmed, while the other 2 of the predefined hypotheses were confirmed for the construct validity. Factor analysis

Table 1. *Patient characteristics*

	Dysfunctional Voiding Group		Reference Group		
	Pts	Parents	Children	Parents	p Value
Mean yrs age (range)	9 (7–10)		10 (8–12)		0.05 (Mann-Whitney U test)
No. female/total No. (%)	24/50 (48)		28/60 (47)		0.45 (chi-square test)
Median (p)VSSDES score (IQR):					
Baseline	17 (13–21)	17 (13–20)	6 (4–8)	5 (3–7)	<0.001 (Mann-Whitney U test)
Retest	15 (10–20)	16 (12–19)			
p Value (Wilcoxon signed rank test)	0.005	0.99			
Median (p)PiNQ score (IQR):					
Baseline	21 (15–31)	20 (15–29)			
Retest	19 (12–26)	21 (12–28)			
p Value (Wilcoxon signed rank test)	0.01	0.63			

A total of 32 patients and parents were available for retesting.

Table 2. *Interrater reliability between child and proxy scores for VSSDES*

	No.	ICC (95% CI)
Patients:		
Younger than 9 yrs	23	0.81 (0.61–0.92)
Older than 9 yrs	26	0.44 (0.07–0.70)
Total/av	49	0.63 (0.41–0.77)
References:		
Younger than 9 yrs	18	0.72 (0.39–0.89)
Older than 9 yrs	42	0.68 (0.48–0.81)
Total/av	60	0.72 (0.57–0.82)
Overall	109	0.85 (0.79–0.89)

demonstrated the 4 previously identified dimensions, incontinence, urgency, obstructive symptoms and fecal complaints, explaining 60% of the variance.

ROC analysis revealed a cutoff score of 11 for the VSSDES with a sensitivity of 92% and a specificity of 85% (AUC 0.95, 95% CI 0.91–0.99). For the pVSSDES a cutoff score of 9 with 94% sensitivity and 91% specificity (AUC 0.98, 95% CI 0.95–1.00) was found (see figure). [F1]

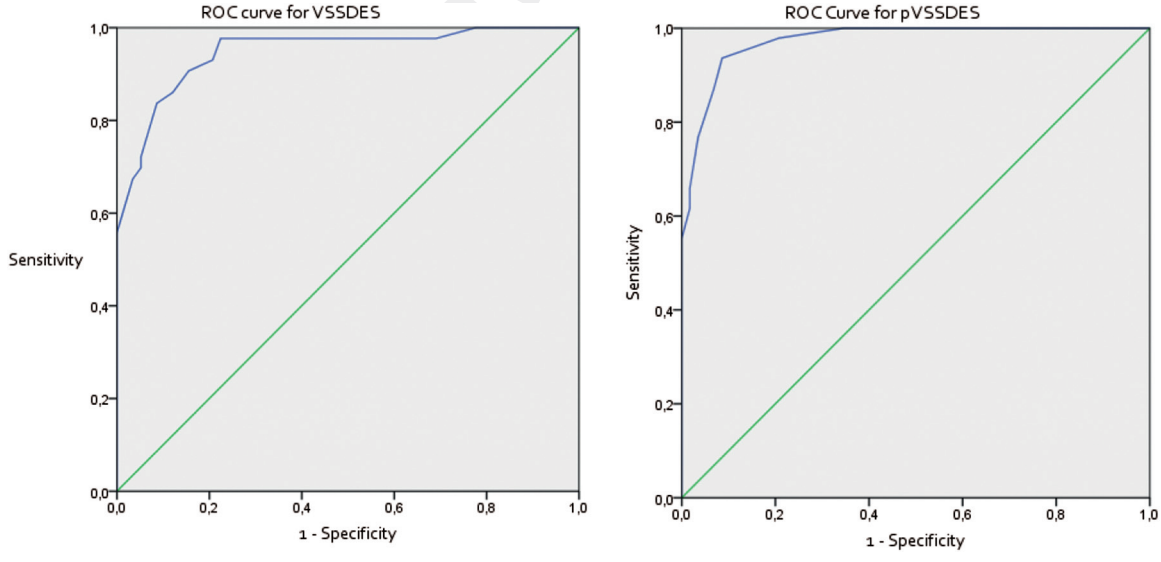
DISCUSSION

The primary aim of this study was to translate and cross-culturally validate the Dutch language version of the VSSDES in patients with DV. Reliability and validity analyses showed moderate to adequate results, comparable to the original development study.[10]

The patient group reported significantly higher scores than the reference group, indicating a discriminative ability and possible diagnostic value of the VSSDES for patients with DV. A moderate internal consistency was found with a Cronbach alpha of 0.55 in the patient group. This was slightly higher than the Cronbach alpha (0.45) described in the original validation study.[10] In the Chinese validation of a different measure for DV a moderate internal consistency (Cronbach alpha 0.45) was found as well.[18] This lower Cronbach alpha can be explained by the range of aspects that are associated with DV. The current factor analysis confirmed the 4 different factors previously identified and could thereby justify the heterogeneity of DV as a reason for the moderate internal consistency.

Test-retest reliability (ICC 0.41) was less in the current study than in the original validation study. The test-retest period was a median of 15 days, compared to 1 week in the original validation study.[10] Although no official treatment was initiated during the retest period, general recommendations regarding voiding were probably provided during the first visit, potentially resulting in improvement of DV symptoms. This assumption is supported by the significant improvement of VSSDES scores at the retest found in this study. When selecting only those patients whose test-retest period was 1 week, the Dutch version of the VSSDES demonstrated adequate test-retest reliability (ICC 0.79-0.94).

VSSDES scores of children and their parents were highly correlated in all groups. However, in patients 9 years or older the scores on the VSSDES were only slightly correlated with those of their parents. In clinical practice it is important to be aware of this difference in perception between children and parents. Younger children are usually more dependent on their parents regarding the specifics of daily life than older children. In children



ROC curves for VSSDES and pVSSDES.

up to 9 years old the parent scores are likely representative of the actual symptoms, while in older patients separate patient scores could provide additional information regarding the presence of symptoms. This disparity between patient and parent reporting of severity of symptoms has been observed previously,[19] and a similar discrepancy has been found in assessment of QoL.[20,21]

In the present study only a slight correlation between the VSSDES and PiNQ was found, while we found no correlation at all between the pVSSDES and pPiNQ. This observation may be explained by the potential tendency of parents to underestimate the impact of symptoms on QoL of their children.[22] However, this low correlation does not indicate a low validity. The PiNQ was chosen as a substitute since a gold standard is lacking for assessment of DV. The PiNQ focuses on the impact of symptoms of DV on QoL and not on the actual severity of symptoms. We hypothesized that a correlation between the VSSDES and the PiNQ would be found, although we observed only a slight correlation for the PiNQ and no correlation for the pPiNQ.

ROC analysis to differentiate between the patient and reference groups confirmed the same cutoff value of 11 for the child scores found in the original development and validation study.[10] For the parent scores a lower cutoff of 9 was observed. These findings could be explained by the difference in score distribution between parents and patients in the reference group. In the reference group parent scores were slightly lower than subject scores, resulting in a lower cutoff score for DV in parents.

Strengths of this study include use of standardized measurement properties to evaluate the reliability and validity of the Dutch VSSDES.[14] Furthermore, data for the reference group confirm the potential of the VSSDES to differentiate between patients with and without DV. In addition, the sample of patients was recruited in different hospital settings, which makes the results of this study more generalizable in clinical practice.

Limitations of the study include the unknown response rate. Treating physicians were asked to recruit all consecutive eligible patients. However, if patients did not participate, it is unclear whether they were unwilling to or had not been asked. Also a modest Cronbach alpha was observed. However, this finding does not immediately imply low reliability because of the heterogeneous character of DV. Furthermore, although the sample size was adequate,[14] results could have been influenced by the relatively small size of the patient group. A responsiveness analysis was not performed.

CONCLUSIONS

We have fashioned a reliable and valid Dutch questionnaire to more systematically evaluate the symptoms of patients with DV. The Dutch VSSDES will support physicians in their assessment during clinical work but will also support standardized reporting of outcomes of clinical research for functional pediatric urology. In children older than 9 years using the VSSDES scores in addition to the parent scores could provide additional information regarding symptoms. Future research should focus on the responsiveness and clinical applicability of the Dutch VSSDES.

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