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Health-related quality of life in patients with cleft palate: Validity and reliability of the VPI Effects on Life Outcomes (VELO) questionnaire translated to Dutch

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ABSTRACT

Background: Disease-specific health-related quality of life (HRQOL) questionnaires provide the clinician with important information regarding the impact of the disease on functioning and well-being. For patients with velopharyngeal insufficiency (VPI), the VPI Effects on Life Outcomes (VELO) questionnaire was developed and validated in English by Skirko et al. (2012). However, a valid and reliable Dutch translation of this questionnaire is not available yet.

Methods: The English questionnaire was translated to Dutch following a forward-backward translation procedure. A linguistic validation and the evaluation of the internal consistency (Cronbach's α) of this Dutch version were performed based on the responses of 39 parents of patients with cleft (lip and) palate (mean age: 6.8 years) (parent report) and the responses of 14 patients older than 8 years (mean age: 9.5 years) (child report). Additionally, the concurrent validity was assessed by comparing the scores on the parent report to those on the pediatric voice handicap index. Furthermore, the validity of the parent proxy assessment and the relationship between age and responses on the VELO questionnaire were investigated. Based on the responses of an age and gender matched control group without cleft palate, the discriminant validity was evaluated.

Results: The parent report was easy to complete for all parents. Nine of the fourteen (64%) patients were able to complete the child report independently. The median scores on the parent report and the child report were 82.7 and 95.1 respectively. The patient group had a significantly worse perception of HRQOL compared to the control group ($p < 0.001$; $p = 0.029$). There were no significant differences between the responses of the parent and their child's ($p = 0.345$). A significant positive correlation was found between the score on the parent report and the age of the patients ($p = 0.001$). Furthermore, a significant negative correlation was found between the parent report and the P-VHI ($p < 0.001$). Cronbach's α was 0.955 and 0.817 for the parent report and the child report respectively.

Conclusion: The Dutch VELO questionnaire is a valid, reliable and user-friendly tool that provides important information about HRQOL in patients with cleft (lip and) palate.

1. Introduction

Quality of life is a well-known concept describing a person's well-being, and is considered to be an important parameter to assess treatment outcomes [1]. More specifically, health-related quality of life (HRQOL) is a commonly used approach to describe a patient's perception of the effect his/her health status has on functioning and well-being [2–4]. HRQOL instruments are characterized by their subjective and multidimensional nature, addressing minimally physical, mental and social domains of health

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[3,5–7]. These instruments can be either generic or disease-specific. Generic instruments facilitate the comparison between populations with and without a disease. Disease-specific instruments on the other hand obtain information related to a certain pathology [4].

With an incidence of approximately 1 in 1000 live births, clefts of the (lip and) palate (C(L)P) are one of the most common congenital deformities [8]. This disruption of the facial structure causes an abnormal velopharyngeal anatomy, frequently resulting in the inability to accomplish closure of the velopharyngeal valve, even following primary palatal closure [9], which is referred to as velopharyngeal insufficiency (VPI) [10]. Clefts of the (lip and) palate are the most common cause of VPI. However, other congenital causes such as velar dysplasia [11] and acquired conditions such as VPI following adenoidectomy [12] have been described. Adequate velopharyngeal function is important for the correct production of speech sounds and swallowing. Consequently, VPI can affect these acts and inherently psychosocial aspects of life [13,14]. Considering this distinctive impact of VPI, a disease-specific instrument could provide insight in the degree to which a patient's functioning and well-being is affected by VPI [14]. Furthermore, a valid and reliable VPI-specific HRQOL instrument should be able to detect changes in HRQOL [15].

Such a disease-specific instrument for patients with VPI was developed by Barr et al. [14]. Their Velopharyngeal Insufficiency Quality-of-Life (VPIQL) instrument consisted of 48 items which were retained following focus groups, including patients and their parents, and clinician's panels. However, Skirko et al. [16] found that 22 items of this questionnaire were redundant. These redundant items were identified following statistical analysis including the detection of floor and ceiling effects based on the endorsement frequency, item-total correlation and item-item correlation. Subsequently, a panel of clinicians, two pediatric otolaryngologists and one speech-language pathologist, decided to eliminate these items.

Elimination of these 22 items resulted in the VPI Effects on Life Outcomes (VELO) instrument [16]. The VELO questionnaire consists of a component for the parent (parent report) as well as for the patient (child report), with each item being scored on a Likert-type scale, ranging from zero (never) to four (almost always). The parent report comprises 26 items addressing six domains: speech limitation (7 items), swallowing problems (3 items), situational difficulty (5 items), emotional impact (4 items), perception by others (4 items) and caregiver impact (3 items). The 23-item child report addresses the same domains, except caregiver impact. Similar to the score on the PedsQL$^{4.0}$ [17], a generic HRQOL questionnaire for the pediatric population, the total scores on the VELO questionnaire and on the subscales range from 0 to 100, with 100 representing the highest quality of life [16]. A first evaluation of the reliability and validity of the VELO questionnaire showed excellent internal consistency, discriminant validity and concurrent validity with the PedsQL$^{4.0}$ [16]. Importantly, the readability of the VELO questionnaire was also evaluated and improved based on the results of the Flesch-Kincaide Grade Level [16,18]. A subsequent study by Skirko et al. [19] demonstrated concurrent validity of the VELO instrument with the Pediatric Voice Outcomes Survey [20], the Pediatric Voice Related Quality of Life [21], and a combined visual analogue scale evaluating speech, swallowing, and situational and social interactions. Furthermore, excellent test-retest reliability, anatomic construct validity and responsiveness of the VELO instrument to change of the quality of life three months after treatment were found. Finally, a recent study also showed sensitivity of the VELO questionnaire to clinically important VPI-specific quality of life improvements following Furlow palatoplasty and sphincter palatoplasty [22].

Although the VELO questionnaire has shown to be a valid and reliable instrument to measure HRQOL in children with VPI, this instrument cannot be used in non-English speaking populations. Therefore, a careful translation and a rigorous testing of the validity and reliability in the specific cultural context is required [23,24]. Hence, the purpose of the current study was to translate the VELO questionnaire to Dutch and to analyze the validity and reliability of the translated version based on the responses of patients with C(L)P and their parents, as this congenital malformation is the leading cause of VPI [25].

2. Methods

This study was approved by the ethical committee of the Ghent University Hospital (2016/0338). All patients and their parents participated voluntarily and signed an informed consent.

2.1. Translation of the VELO questionnaire to Dutch

A forward-backward translation procedure was conducted by the authors, following the principles of good practice described by the ISPOR Task Force for Translation and Cultural Adaptation [26]. The forward translation from English to Dutch was conducted independently by two Dutch speaking researchers with a professional proficiency in English. They also had clinical and research experience in patients with C(L)P. Thereafter, both Dutch versions were reconciled until a consensus was reached. The backward translation from Dutch to English was conducted by an English native speaker. Finally, following review of the backward translation by comparing this translation with the original instrument, a definite Dutch version was constructed. This procedure was followed for both the parent report and the child report. During the translation process, special attention was given to the wording to increase readability. In order to evaluate and improve the performance of the questionnaire, all parents participating in the current study were asked to evaluate the clarity and readability of the items. Furthermore, the parents were asked to indicate whether their child was able to complete the questionnaire independently and if not, what was unclear.

2.2. Subjects and data collection

Thirty-nine subjects with a history of C(L)P aged between 3 and 12 years (*Mean (M)* = 6.8 years, *Standard Deviation (SD)* = 2.41) were enrolled between July 2016 and November 2016. Seven patients had a bilateral cleft lip and palate (BCLP), 16 patients presented with a unilateral cleft lip and palate (UCLP) and 16 patients had a cleft palate only (CP). All patients were followed by the multidisciplinary craniofacial team of the Ghent University Hospital. They all had Dutch as their mother tongue, as well as their parents. Indications of VPI were not taken into account for the inclusion of the participants. Patients with a syndrome, a moderate or severe hearing loss or severe cognitive impairment were excluded. This information was retrieved from the patient's medical records.

The control group was recruited by convenience and snowball sampling, and consisted of participants without cleft palate or any other craniofacial malformation, cognitive impairment, moderate or severe hearing loss, neurological deficit, or previously diagnosed speech or language disorder. The absence of these criteria was evaluated based on the subjective report of the parents. The participants of the control group were matched with the patients for gender and age. Both the experimental group and the control group consisted of 13 boys and 26 girls. The mean age of the control group was 6.7 years (SD = 2.38), which was not significantly different from the experimental group ($t(76) = 0.24$, $p = 0.814$).

Similar to the procedure described by Skirko et al. [19], only the