

Application of a new patient-reported outcome measure in orofacial clefts: An exploratory study in two countries

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SUMMARY

Patients with repaired cleft lip and/or palate may vary in satisfaction with their treatment. The exploratory study investigated the satisfaction of patients with orofacial clefts, and their parents with cleft treatment outcomes using the Cleft Hearing, Appearance and Speech Questionnaire (CHASQ). The study included 29 Vietnamese and 27 Estonian patients aged ≥ 7 years with repaired cleft lip and/or palate. The CHASQ was translated into Vietnamese and Estonian. The questionnaire was completed independently by patients and their parents. There were nine items in the CHASQ associated with the cleft (Factor 1) and six items less associated with the cleft (Factor 2). Significant moderate positive correlations were related to Factor 1. Vietnamese patients self-rated lower than Estonian patients in most of the items except speech. The agreement between patients and parents varied from low to moderate positive correlations in the features associated with the cleft, and mainly in the Vietnam sample. Vietnamese patients were less satisfied than Estonian patients. CHASQ is an easy tool to evaluate patients' satisfaction with hearing, appearance, and speech.

Key words: cleft lip; cleft palate; patient satisfaction; speech; treatment outcome.

INTRODUCTION

Cleft lip and/or palate (CL/P) includes cleft lip with or without cleft alveolus (CL \pm A), cleft lip and palate (CLP), and cleft palate (CP) (1). Clefts involving the lip and/or alveolus (i.e. CL \pm A, CLP) affect appearance and occlusion (2). Clefts involving the palate (i.e. CP, CLP) affect feeding, hearing, and speech (2, 3). Patients with CL/P, therefore, often undergo multiple treatments to achieve the best functional and esthetic outcomes from birth to adulthood.

The treatment outcomes could be assessed by doctors, parents, or patients. Since patients are considering as the center of any healthcare system, the provision of health care is shifting from a traditional paternalistic approach to a patient-centered care approach (4). Thus, it is crucial to measure patient-reported outcomes as a key determinant of treatment success rather than other outcomes like parent-reported or doctor-reported (5). Patient-reported outcomes help understand the current treatment from the patients' viewpoint (5) and avoid observer bias (5, 6).

There is a lack of validated cleft-specific instruments to gather patient-reported outcomes in practice (7-9). Responding to the call for a cleft-specific patient-reported outcome measure, the Cleft Hearing, Appearance and Speech Questionnaire (CHASQ) was developed. The CHASQ (/ˌtʃæsˈkjuː/) was designed and validated specifically for use with patients with CL/P and parents of children with CL/P. It measures patients' and parents' satisfaction with three domains: appearance, speech, and hearing. It has been used routinely in the UK for audit purposes since it was developed (10).

Due to the fact that most of the available patient-reported outcome measures were developed in the English language, there is a need to translate and adapt the instruments for use in other countries to increase the international involvement (11). Moreover, cleft treatment protocols and timings vary greatly within and between countries. Multinational as well as multicenter studies, therefore, are becoming more common to compare the cleft outcomes across countries. Consequently, under the umbrella of the COST Action IS1210 Appearance Matters in 2014, the CHASQ was translated and piloted in some European countries and in Vietnam. The goal was to implement the CHASQ in practice internationally.

This exploratory study aimed to: 1) develop the Vietnamese and Estonian versions of the CHASQ,

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2) explore the satisfaction of patients with CL/P and their parents with the outcomes of the cleft treatment in Vietnam and Estonia based on the CHASQ, and 3) investigate patient-parent agreement on the satisfaction of the treatment.

MATERIALS AND METHODS

The Cleft Hearing, Appearance and Speech Questionnaire (CHASQ)

The CHASQ was developed by the Cleft Psychology Special Interest Group, Craniofacial Society of Great Britain and Ireland specifically for patients with facial disfigurement (12). The CHASQ is a 15-item questionnaire with the answers being given on an 11-point rating scale ranging from 0 (“very unhappy”, “not at all good-looking”, or “very noticeable”) to 10 (“very happy”, “very good-looking”, or “not at all noticeable”). The score from 6 to 10 is within the norm for the cohort; from 1 to 5 is less satisfied than the norm; and 0 is much less satisfied than the norm (10).

The 15 items are: face, whole appearance, side view/profile, good-looking, nose, lips, chin, teeth, cheeks, hair, ears, eyes, speech, hearing, and noticeability. These items are classified into two factors using an exploratory factor analysis with promax rotation. Factor 1 is features that are associated with having been born with a cleft. Factor 1 includes nine items: face, whole appearance, side view/profile, good-looking, nose, lips, teeth, speech, and noticeability. Factor 2 is features that are less associated with having been born with a cleft. Factor 2 includes the other six items: chin, cheeks, hair, ears, eyes, and hearing. The factors have a high internal consistency: Factor 1 ($\alpha=0.9$) and Factor 2 ($\alpha=0.83$) (10).

The linguistic validation process of the CHASQ

The validation process followed an established procedure that consisted of three stages: forward translation, backward translation, and patient testing (13). The linguistic validation process was to ensure semantic equivalence between a source language and a target languages (11). The source language of the CHASQ was English. The target languages were Vietnamese and Estonian.

For the Vietnamese version, in the first stage, three translators who were native Vietnamese speakers and bilingual in English translated the questionnaire into Vietnamese independently. The translators and the author (V.T.N) discussed and agreed on the reconciliation version (the first version). In the second stage, the translators were Vietnamese postgraduate students studying in English-speaking

countries (the USA and Australia) for at least two years. They translated the first version into English. The translators and the author (V.T.N) discussed to detect any misunderstandings, mistranslations, or inaccuracies. The comparison resulted in changes in the first version; the second version was produced. The second version was tested on five Vietnamese patients with a cleft. The author (V.T.N) asked the patients if they had any difficulties in understanding. The third version (the final version) was released based on the feedback from the patients (Fig. 1).

For the Estonian version, a similar process was done. In the first stage, three translators translated the questionnaire into Estonian independently. The translators and the author (T.J.) produced the first version after discussion. In the second stage, one Estonian translator who can speak English fluently translated the first version into English. The second version was produced after discussion between the translator and the author (T.J.). The second version was tested on five Estonian patients with a cleft. The final version was released after corrections based on the feedback from the patients (Fig. 2).

Participants

The study was approved by the Institutional Ethics Committees of Hue University of Medicine and Pharmacy (24 December 2015) and the University of Tartu (reference number: 278T-1). The written consent was obtained from the patients or parents of patients who were 18 years or younger.

The exploratory study took place at Hue University Hospital (Hue, Vietnam) and Unimed Clinic (Tartu, Estonia). Patients with repaired CL/P who visited the hospital/clinic were invited for the study. Patients aged from 7 years were eligible for the study. The lower age limit of 7 years was chosen because participants younger than this would have difficulty completing the questionnaire. Only pairs which both patient and parent completed the questionnaire were included. Twenty-nine Vietnamese patients (7–28 years old; median age 15 years old) and 27 Estonian patients (8–19 years old; median age 12.3 years old) agreed to participate in the study. The patients' cleft treatment history is demonstrated in Table 1.

Procedure

The procedure was the same for both Vietnam and Estonia samples. The participants (both the patients with CL/P and their parents) completed the CHASQ by themselves. The parents and patients were in different rooms when completing the questionnaire. Examiners could explain the questionnaire to the participants if the participants did not

Wilk test for normality showed that the data was not normally distributed in many items ($p < 0.05$); therefore, nonparametric tests were used. A Mann-Whitney U test was used to compare the differences in the ratings between Vietnamese and Estonian patients. A Wilcoxon signed-rank test was conducted to compare the differences in the satisfaction between patients and their parents. A Spearman correlation coefficient was used to test the level of agreement between patients and their parents.

RESULTS

Satisfaction of patients

Patients scored their satisfaction above 5.0 for most of the items of the CHASQ. Considering items in Factor 1, Vietnamese patients were least satisfied with lips and teeth; Estonian patients were least satisfied with noticeability.

Mann-Whitney U tests showed that there were significant differences in the satisfaction between Vietnamese and Estonian patients with all items in Factor 1 and Factor 2 ($p < 0.05$), except for speech ($p > 0.05$) (Table 2).

Satisfaction of parents

While Vietnamese parents rated their satisfaction higher than their children, the satisfaction of Estonian parents was lower than their children. The Wilcoxon signed rank test showed Vietnamese parents rated significantly higher than their children in eight items: face, whole appearance, side view/profile, good-looking, lips, chin, teeth, and ears ($p < 0.05$) (Fig 3A). In contrast, Estonian parents rated their satisfaction significantly lower than their children in four items: lips, teeth, speech, and noticeability ($p < 0.05$) (Fig 3B).

Patient-parent agreement

A Spearman’s correlation was run to determine the relationship between patients and their parents’ ratings. There were no significant high or very high positive correlations between patients and parents in both Vietnam and Estonia samples. Specifically, in Vietnam, moderate and significant correlations were observed in respect of the items: face, nose, lips, teeth, and speech. In Estonia, fewer correlations were identified. Moderate and significant correla-

Table 1. Cleft treatment protocol (timing and surgical method) in Vietnam and Estonia

	Vietnam	Estonia
Lip repair	3–6 months; Millard method	4–5 months; Millard method
Palate repair	18–24 months; modified von Langenbeck method	9–14 months; Veau-Wardill-Kilner pushback method
Alveolar bone graft	None	9–11 years depending from canine position; bone from iliac crest
Speech therapy	None	3 years
Orthodontics	None	5–6 years
Psychological counselling	None	None

Table 2. Median scores of each item of the CHASQ scored by Vietnamese and Estonia patients. Comparison between Vietnamese and Estonian patients’ ratings

Items	Vietnam (n=29)	Estonia (n=27)	p-value*
Factor 1			
Face	7	10	0.004
Whole appearance	6	10	< 0.001
Side view/Profile	5	10	0.001
Good-looking	5	9	< 0.001
Nose	7	9	0.007
Lips	5	10	< 0.001
Teeth	4	9	< 0.001
Speech	8	9	0.093
Noticeability	5	8	0.012
Factor 2			
Chin	8	10	0.001
Cheeks	8	10	0.001
Hair	9	10	0.004
Ears	8	10	< 0.001
Eyes	9	10	0.008
Hearing	9	10	0.021

* The Mann-Whitney U test was used to compare between Vietnamese and Estonian samples

tions were found in the item of nose.

DISCUSSION

To our best knowledge, this is the first study on the satisfaction of patients and parents with appearance, speech, and hearing using the CHASQ in Vietnam. The CHASQ was a helpful questionnaire for screening patients’ needs in a relatively short time and shifted the medical/aesthetic focus towards patients (14).

Satisfaction of patients and parents

The results showed that patients and their

parents were satisfied with their appearance, speech, and hearing. Interestingly, although Vietnamese patients with CL/P in our study did not receive speech therapy, they were not unsatisfied with their speech. The patients might learn how to adjust their speech, or have become satisfied with their speech condition, or were not aware that their speech could be better.

Vietnamese patients self-rated significantly lower than Estonian patients in almost every item. Vietnamese patients had lower satisfaction could be because of low self-esteem, less information about their cleft, and their perception of the standard of beauty. It was documented in the literature that East

Asian people reported lower levels of self-esteem than those from Western countries (15, 16). The later the patients learned about their CL/P, the less they were satisfied with their condition and the lower self-esteem they had (17).

Patients were least satisfied with teeth in both samples, but more drastic in the Vietnam sample. It might be because patients in Vietnam have not received any orthodontic treatments and had more visible caries in front teeth than patients in Estonia. The teeth were not well aligned due to the lack of orthodontic treatment could be the reason for low satisfaction (18). A review has shown that patients with a cleft had a higher risk of caries than noncleft individuals (19).

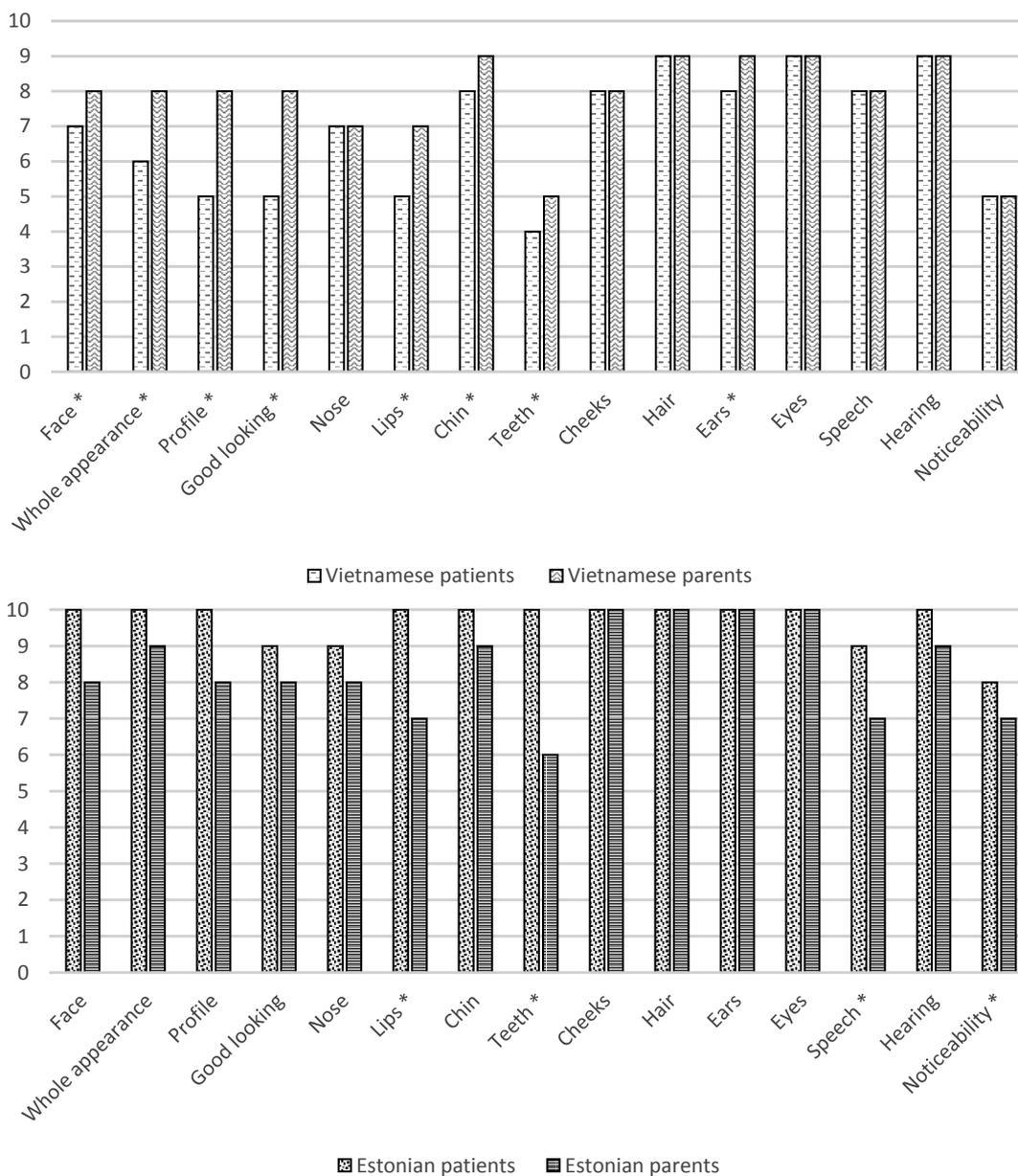


Table 3. Median scores of each item of the CHASQ scored by (A) Vietnamese patients and their parents; (B) Estonian patients and their parents. The Wilcoxon test was used to compare the score between the patients and their parents.

There were some differences between Vietnamese patient and parent ratings for features related to appearance. Parents can be more honest to disclose their concerns than their children. However, in case the parents thought they were not in the position to complain about the outcomes since they had received free health care, they might exaggerate their satisfaction or set their satisfaction lower than prior expectations (20). Thus, it is important to ask a child's opinion of the treatment outcome independently from the parents, and treatment planning should not be based merely on parents' opinion. Less differences were observed between Estonian children and parents compared with the Vietnam sample. The difference between the two

samples could be because of cultural background, external environment, and the level of understanding about cleft between Vietnamese and Estonian patients/parents.

Patient-parent agreement on the satisfaction

One of the aims of our study was to investigate the patient-parent agreement on the satisfaction using the CHASQ. Significant moderate correlations between patients and parents were found because of a similar experience related to the cleft of the patients and parents. More correlations were observed in the Vietnam sample than in the Estonia sample. Earlier studies also reported a low/fair to a moderate interrater agreement between patients and their parents in different measures; however, the level of agreement was incomparable because of different statistical analysis or interpretation of the level (2, 18, 21).

Clinical implications

Items of the CHASQ are related to facial features that play an important role in evaluating facial appearance in patients with CL/P. They can be used to determine patients' and parents' satisfaction with the clinical outcome of cleft treatment, for example, before and after secondary surgeries or speech therapy. Clinicians can determine any significant changes in their satisfaction with features related to appearance, speech and hearing. The CHASQ can also be administered to assist therapeutic interventions as an assessment tool or outcome measure for patients, and therefore has been used in studies as a part of a test battery when examined the impact of satisfaction on psychological factors such as mood, confidence, self-esteem, and quality of life (10).

Limitations and future direction

The limitation of the study and directions of

future research should be addressed. One of the limitations was a small sample size. For exploratory and pilot studies, a sample size of 30 subjects (or an estimate of 24 to 36) was suggested for some practical advantages, such as minimum cost, simplicity, easy calculation, and the ability to test hypotheses (22).

When informed about the purpose of the study, patients, as well as their parents, were reluctant to participate. The rationales of not participating were an expectation of further treatments involved rather than just inspecting the satisfaction, far distance from the center where the study was conducted, and the non-covered travel expenses.

There is a need that psychosocial assessment should be incorporated into the treatment protocol and assessed regularly. Especially, since satisfaction with appearance significantly correlates with self-reported psychosocial functioning (23).

CONCLUSIONS

The validated CHASQ was available in the Vietnamese and Estonian language, and ready for use. Patients and parents were satisfied with the outcomes of the cleft treatment. The correlations between patients' and parents' ratings ranged from low to moderate positive correlations in the features that were appearance-related and cleft-associated, and mostly in the Vietnam sample. Vietnamese patients were less satisfied with features associated with their appearance and hearing than Estonian patients.

CONFLICTS OF INTEREST

The authors state no conflict of interest.

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