Original paper

Evaluation of the effect of palatoplasty on the quality of life and speech outcomes in patients with velocardiofacial syndrome

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Conflict of interest
None declared

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Abstract

Background. Children diagnosed with velocardiofacial syndrome (VCFS) suffer from various disabilities. Palatal abnormalities, as well as speech and language impairment, adversely affect a child's quality of life (QoL) and are some of the most distressing aspects for the parents of these children.

Objectives. The present study aimed to explore the effect of palatoplasty on the health-related quality of life (HRQoL) and speech outcomes in children with VCFS.

Material and methods. The study recruited 20 patients ($N = 20$) with VCFS and connected speech, aged 3 years or older, having either undiagnosed submucous cleft palate (SMCP) or velopharyngeal insufficiency (VPI), and requiring primary cleft palate surgery or revision surgery. Speech assessment was conducted prior to palatoplasty and 6 months after the surgery. Intelligibility and hypernasality were evaluated using the Cleft Audit Protocol for Speech – Augmented (CAPS-A). The parent proxy-report form of the Pediatric Quality of Life Inventory (PedsQL™) was used to evaluate and compare the HRQoL of the VCFS patients before and after palatoplasty.

Results. Significant improvement in the HRQoL scores was achieved after the surgery across all domains (physical, emotional, social, and school functioning), especially in the emotional and social dimensions ($p < 0.000$). The post-operative speech assessment based on CAPS-A demonstrated improvement in speech intelligibility and hypernasality in the majority of patients.

Conclusions. Given that children with VCFS face various medical and social problems, suitable palatal interventions are beneficial, improving both the speech ability and QoL of these children.

Keywords: quality of life, DiGeorge syndrome, cleft palate

Introduction

Velocardiofacial syndrome (VCFS) is one of the most common examples of an autosomal dominant disorder caused by a micro-deletion of chromosome 22q11.2, affecting nearly 1 in 3,000 children.[1] The syndrome is diagnosed with an accurate and reliable special blood test called fluorescent in situ hybridization (FISH).[2] Children with VCFS display many problems that can be challenging to their parents and family members. Typical facial characteristics of the syndrome are posteriorly rotated ears or simple helices as external ear phenotypes. The permanent hearing loss associated with palatal anomalies has also been reported.[3] Of the 180 possible physical symptoms, palatal abnormalities are present in 69–100% of the affected children, with 9–11% having cleft palate, 5–16% having submucous cleft palate (SMCP) and 27–92% suffering from velopharyngeal insufficiency (VPI). Palatal anomalies, as the most prevalent features, result in a decreased physical function affecting the feeding abilities (i.e., growth retardation), a poor functional status and speech difficulties.[4] Effective communication and social interactions are directly associated with an individual's social well-being. Therefore, speech and language problems may adversely affect the quality of life (QoL) in children with VCFS, posing a major concern to their parents.[5,6] A previous study reported that a large proportion of people with a voice disorder experienced a decrease in QoL.[7]

Patients with chromosome 22q11.2 deletion syndrome (22qDS) commonly present with a large central velopharyngeal gap in the setting of poor velar and pharyngeal wall motion. The presence of a velopharyngeal gap can be identified based on a listener's judgment, oral examinations and sound production tests.[8] The Cleft Audit Protocol for Speech – Augmented (CAPS-A) is a validated and reliable tool to evaluate the speech of patients with cleft palate and VPI.[9,10] Due to advances in medicine, pediatric healthcare expands beyond the physical health of a child to include their health-related quality of life (HRQoL).[11,12] Researchers agree that QoL is a multi-dimensional concept that encompasses physical, psychological, social, and spiritual aspects.[13] The health-related quality of life can help translate a patient's experience of illness into quantifiable outcomes. Clinicians and researchers can determine the effectiveness of interventions to improve QoL in individuals with acute or chronic health problems. The quality of life in children with chronic conditions, such as VCFS, is affected by complex and dynamic interactions between various factors. Interactions between physical, emotional, social, and school functioning significantly influence both the overall well-being and QoL of a child.[14,15] The Pediatric Quality of Life Inventory (PedsQL™) offers a modular approach to measure HRQoL in healthy children and adolescents, and in those with acute or chronic health conditions.[16,17]

Since VCFS patients suffer from a wide range of problems, their speech impairment is often ignored in the light of other possible disabilities. Surgical protocols are contradictory, which complicates the treatment of VCFS patients. However, appropriate palatal surgery can be effective in enhancing patients' QoL. Hence, the present study aimed to explore the post-operative effect of palatoplasty on HRQoL and speech outcomes in children with VCFS.

Subjects and methods

The present study was conducted over 5 years (2014–2019) at the Cleft Lip and Palate Center of Shiraz University of Medical Sciences, Iran. The target population included patients with VCFS and those suspected of having the syndrome who were referred to the center. The FISH test was performed to confirm the diagnosis of VCFS in the suspected patients. The inclusion criteria were children with VCFS and connected speech, aged 3 years or older, having either undiagnosed SMCP or VPI, and requiring primary or revision repair. Of the initial 45 eligible candidates, 20 patients met the inclusion criteria and were recruited in the study. Prior to speech assessment, the parents of the children were informed about the research goals, the test procedure, and what was expected of them during the assessment. Written informed consent was obtained from the parents of the patients, and the anonymity of the children was guaranteed. The study protocol was approved by the institutional Ethics Committee at Shiraz University of Medical Sciences (IR.SUMS.REC.1398.535).

Speech assessment was carried out in 2 stages – prior to palatoplasty and 6 months after the procedure. In line with a previous study,[18] a digital voice recorder (DM-3; Olympus, Tokyo, Japan) and a camera (DCR-SR62E; Sony, Tokyo, Japan) were used to record the voice and image of the participants. The voice recorder was positioned at a distance of 15 cm from the patient's mouth, and the recordings were made in a quiet and well-prepared environment. Since the recordings before and after palatoplasty were taken by a third party, blind data analysis was ensured. The same person randomly organized, coded and stored the recordings on a digital optical disc. Subsequently, an independent speech–language pathologist, specialized in orofacial cleft malformation, speech intelligibility and hypernasality in such patients, analyzed the data from the 20 participants with the use of CAPS-A. To ensure accuracy, the data from 10 participants was randomly selected from the same dataset, re-analyzed and compared with the initial analysis.

In the present study, we evaluated the intelligibility and hypernasality parameters of CAPS-A. The assessment involves data interpretation based on colors; each color represents a level of speech difficulty: green – normal; yellow – needs further investigation and improvement; and red – unsatisfactory.[10] Intelligibility is the degree to

which speech can be understood by an unknown listener. The judgement is based on a short sample of conversational speech, and scoring comprises 5 grades, namely 0 (normal), 1 (different from other children's speech, but not enough to cause comments), 2 (different enough to provoke comments, but still intelligible), 3 (barely understandable to strangers), and 4 (impossible to understand). Hypernasality is associated with an abnormal increase in the nasal resonance of the voice during speech production. The tone of the voice is most perceptible with vowels and voiced consonants. Hypernasality is also scored on a 5-point scale: 0 (absent); 1 (borderline/minimal); 2 (mild); 3 (moderate); and 4 (severe).[10]

The degree of palate movement and the severity of VPI were assessed based on the analysis of the gap size, the evaluation of lateral video images and nasal endoscopy. Accordingly, the most appropriate surgical procedure (Sommerlad–Furlow modified palatoplasty under magnification or modified Furlow palatoplasty with posterior pharyngeal flap) was selected for each patient and performed by the same surgeon. Six months after palatoplasty, the same speech assessment was conducted to evaluate the outcome of the surgery. Since the children were too young to respond by themselves to the HRQoL questionnaire, the parent proxy-report form of PedsQL was used to assess and compare HRQoL in the VCFS patients before and after palatoplasty. The PedsQL is a 23-item self-report questionnaire that measures the core dimensions of health.[16,17] It includes 4 generic core scales: physical functioning (8 items); emotional functioning (5 items); social functioning (5 items); and school functioning (5 items). In addition, 3 summary scores are incorporated into the scoring procedure: the physical health summary score (8 items); the psychosocial health summary score (15 items); and the total scale score (23 items). All dimensions are evaluated using a 5-point response scale: 0 (never); 1 (almost never); 2 (sometimes); 3 (often); and 4 (almost always). The items are reverse-scored so that lower scores indicate better HRQoL.

Statistical analysis

Descriptive statistics were used to express the intelligibility and hypernasality scores of CAPS-A. The Wilcoxon signed-rank test was used to compare the PedsQL generic core scale scores of the VCFS patients before and after palatoplasty. Spearman's non-parametric test was used to evaluate the correlation between speech assessment and HRQoL. Data from the cognitive assessment of speech and the PedsQL questionnaire was analyzed using the IBM SPSS Statistics for Windows software, v. 23.0 (IBM Corp., Armonk, USA) and the R software for statistical computing and graphics, v. 3.5.1 (https://www.r-project.org). The intraclass correlation coefficient (*ICC*) was used to evaluate the method error. The statistical significance level was set at $p < 0.05$.

Results

A high level of intra-observer agreement was obtained (*ICC* = 0.98). The participants included 12 boys and 8 girls with a mean age of 42 ±4 months. Of the 20 participants, 6 patients underwent primary Sommerlad–Furlow modified palatoplasty, and 14 patients underwent modified Furlow palatoplasty with posterior pharyngeal flap. The mean total scale HRQoL scores in all domains were significantly lower after the surgery. According to the Wilcoxon test, a significant difference was observed in all dimensions before and after the surgery ($p < 0.05$) (Table 1). The CAPS-A pre- and post-operative results showed that 12 patients (60%) had severe hypernasality (grade 4) before the surgery, which decreased to 3 patients (15%) after the surgery. With regard to speech intelligibility before the surgery, 15 patients (75%) scored 4 (impossible to understand), and 5 (25%) scored 3 (barely understandable to strangers). However, after the surgery, the number of patients in the same categories decreased to 1 (5%) and 4 (20%), respectively. Moreover, 15 patients (75%) achieved adequate speech clarity (grades 0, 1 and 2) after the surgery (Table 2). Spearman's non-parametric test showed no significant correlations between the domains of HRQoL, speech clarity and hypernasality before and after palatoplasty (Table 3).

Table 1. Health Related Quality of Life (HRQoL) before and after the surgery (Wilcoxon signed-rank test)

HRQoL domain	Pre-surgery score	Post-surgery score	*p*-value
Physical functioning	8.5500 ±2.98196	8.1000 ±3.00701	0.007*
Emotional functioning	10.8000 ±2.54641	3.8500 ±1.92696	<0.000*
Social functioning	10.6000 ±2.98064	3.7000 ±2.22663	<0.000*
School functioning	6.3500 ±3.19992	5.5000 ±3.15394	0.011*
All domains	36.3000 ±7.88803	21.1500 ±6.19231	<0.000*

Data presented as mean ± standard deviation (*M* ±*SD*).
* statistically significant.

Table 2. Intelligibility and hypernasality parameters of the Cleft Audit Protocol for Speech – Augmented (CAPS-A)

Parameter	Grade	Pre-surgery	Post-surgery
Intelligibility	0	–	1 (5)
	1	–	9 (45)
	2	–	5 (25)
	3	5 (25)	4 (20)
	4	15 (75)	1 (5)
Hypernasality	0	–	1 (5)
	1	–	5 (25)
	2	–	8 (40)
	3	8 (40)	3 (15)
	4	12 (60)	3 (15)

Data presented as number (percentage) (*n* (%)).

Table 3. Correlation between the Health Related Quality of Life (HRQoL) domains and the speech parameters (Spearman's test)

HRQoL domain	Intelligibility	Hypernasality
Physical functioning	0.724	0.278
Emotional functioning	0.674	0.248
Social functioning	0.147	0.353
School functioning	0.606	0.896
All domains	0.310	0.916

Discussion

The results of the present study showed significant improvement in the post-operative HRQoL scores across all domains (physical, emotional, social, and school functioning), especially in the emotional and social dimensions ($p < 0.000$). In addition, the speech assessment based on CAPS-A showed improvement in speech intelligibility and hypernasality. There were no significant correlations between HRQoL and the speech parameters. Our findings are in line with the overall findings of previous studies, suggesting that the complexity of multiple affected systems has a compounding effect on QoL.

Varni et al. evaluated the HRQoL of pediatric patients with chronic conditions, such as diabetes, gastrointestinal conditions, cardiac conditions, asthma, and obesity.[19] They found that their patients had a more impaired overall HRQoL than healthy children.[19] Looman et al. reported that children with VCFS had poorer QoL as compared to healthy children and their peers with a single chronic illness (e.g., diabetes, asthma, cardiac anomalies, and cancer).[20] In addition, the results of the Primary Self-Concept Inventory (PSCI) questionnaire showed that patients with cleft lip and palate had lower scores in 2 sub-scales (social and intellectual self-concept), as well as total scores, when compared to patients with cleft lip or cleft palate only.[19]

Interaction between people is complex, and heavily influenced by appearances and visible differences. It involves communication, self-perception, and how one is perceived by others. Although palatal defects in children are not generally life-threatening, they can be distressing to parents and interfere with feeding. Consequently, the defects can decrease a child's physical functioning, which might limit their ability to participate in recreational activities, thus affecting their social and emotional functioning.[21,22] Damiano et al. evaluated factors that influence the HRQoL of preadolescent children with non-syndromic oral clefts.[23] The authors showed that speech and esthetic concerns were important factors affecting HRQoL in children with oral clefts. Considering the effect of palatoplasty on improving the speech and self-confidence of patients, the surgery was recommended as a method to improve speech outcomes, and therefore, the emotional and social dimensions of HRQoL,[23] which is in line with our findings.

Marcusson et al. evaluated the correlation between facial appearance and QoL in patients who received cleft lip and palate treatment during their childhood.[24] They reported that satisfaction with facial appearance was significantly correlated with better QoL and HRQoL, and dissatisfaction with facial appearance was the most significant predictor of depression in both groups.[24] In our study, we observed improved HRQoL among the children, although no alterations in their facial appearance occurred, as they were only cleft palate patients. This indicates that improvement in speech alone had a significantly positive effect on the HRQoL of these syndromic patients.

Speech development is the most troubling consequence of 22qDS for many parents, since the syndrome causes a delay in acquiring the productive language skills, while the receptive skills are near normal.[25] Learning and cognitive disabilities in patients with VCFS may complicate speech development and therapy.[21]

Children with VCFS exhibit both anatomical and physiological abnormalities in the palate and pharynx, which makes the surgical correction of VPI different from that in non-syndromic children with the repaired cleft palate.[25] Wagner et al. evaluated the outcomes of speech surgery in patients with VCFS, and concluded that the surgical management of VPI in such patients was challenging.[8] Unsuccessful primary surgery poses additional challenges to surgeons during revision surgery, e.g., a scarred pterygopalatine fossa (PPF) donor site, a distorted palatal recipient site, and further medialization of the internal carotid arteries.[8] Posterior pharyngeal fat graft surgery is suggested to improve speech function in patients with VPI. However, surgical protocols are contradictory, which complicates the treatment of VCFS patients.[26] In our study, we used 2 different palatal surgical techniques, depending on each patient's needs. Both techniques yielded positive results, with no clear superiority of one over the other. However, due to the complexity of the syndrome, some patients required the continuation of treatment.

Since the syndrome can affect speech mechanisms, phonation can be aberrant due to a laryngeal web, VPI or vocal cord paralysis. In many cases, phonation may remain abnormal to some extent, even after palatoplasty.[27] Losken et al. evaluated sphincter pharyngoplasty for the management of VPI, and concluded that patients with VCFS were more likely to require pharyngoplasty revision.[28] Similarly, in our study, some patients required pharyngoplasty revision and continual speech therapy, despite significant improvement in hypernasality after the surgery. Overall, the surgical treatment of VPI in patients with VCFS is a challenging task due to anatomical, physiological and neurocognitive abnormalities. However, an appropriate surgical procedure can improve the quality of speech in VCFS patients, thus enhancing their QoL and self-esteem.

Limitations

Due to the rare nature of VCFS, the main limitation of the present study was the small number of participants. It is recommended that future studies involve larger sample sizes and more accurate pre- and post-operative speech evaluation procedures, such as nasometry and video nasopharyngoscopy.

Conclusions

Given that children with VCFS face various medical and social problems, physicians can prioritize their therapeutic needs and improve their QoL by choosing a suitable palatal intervention. The satisfactory outcomes of our post-operative speech assessment support recommending palatoplasty to the parents of such patients as a cost-effective procedure. Furthermore, the PedsQL assessment demonstrated that the intervention was beneficial and improved the HRQoL and speech outcomes of these patients.

Ethics approval and consent to participate

The study protocol was approved by the institutional Ethics Committee at Shiraz University of Medical Sciences, Iran (IR.SUMS.REC.1398.535). Written informed consent was obtained from the parents of the patients, and the anonymity of the children was guaranteed.

Data availability

The datasets generated and/or analyzed during the current study are available from the corresponding author on reasonable request.

Consent for publication

Not applicable.

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