Speech Outcomes and Velopharyngeal Function After Surgical Treatment of Velopharyngeal Insufficiency in Individuals With Signs of Velocardiofacial Syndrome

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Objective: The objective of the study was to analyze if individuals with velocardiofacial syndrome (VCFS) present the same characteristics of speech and velopharyngeal function (VPF) compared with patients with nonsyndromic submucous cleft palate, as well as to compare the effectiveness of palate surgery on the speech function and VPF between groups.

Methods: This was a prospective study performed at the Speech Therapy Sector and Physiology Laboratory, Hospital for Rehabilitation of Craniofacial Anomalies/University of São Paulo.

The procedure performed was primary palatoplasty associated or not to superiorly based pharyngeal flap surgery.

There were 50 patients with velopharyngeal insufficiency: 25 with signals of VCFS (VCFS group) and 25 without syndrome with submucous cleft palate (SMCP group).

The hypernasality was scored by 3 examiners; nasalance was evaluated by nasometry, and VPF was assessed by the size of the velopharyngeal gap on the nasoendoscopy. The evaluations were conducted before and, in average, 18 months after surgery.

Results: Before surgery, the VCFS and SMCP groups presented similar speech function and VPF characteristics in all parameters, with no statistically significant differences. After surgery, there was reduction in the hypernasality, nasalance, and VPF in, respectively, 20%, 31%, and 36% of patients in the VCFS group and in 24%, 30%, and 30% in the SMCP group. Elimination/normalization of variables was obtained in 28%, 19%, and 8% of patients in the VCFS group and 20%, 40%, and 25% in the SMCP group, respectively, for hypernasality, nasalance, and VPF. There was no statistically significant difference between groups.

Conclusions: Patients with VCFS presented similar speech function and VPF characteristics as patients with nonsyndromic SMCP. The surgery for velopharyngeal insufficiency correction was equally effective for the improvement and resolution of speech symptoms and VPF in patients with VCFS compared with the SMCP group.

Key Words: Velocardiofacial syndrome, velopharyngeal insufficiency, speech outcomes, surgery

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Velopharyngeal insufficiency (VPI) is the most frequent symptom among the several clinical signs described for the velocardiofacial syndrome (VCFS), directly interfering with speech production. Its cause may be related to certain structural conditions, such as submucous or occult submucous cleft palate, palatopharyngeal disproportions, hypoplasia or absence of palatal aponeurosis, pharyngeal asymmetry, pharyngeal hypotonia, and hypodynamic velopharynx, and may even persist after surgical correction in cases of overt cleft palate.[1–3]

Some physical and functional factors observed in this population, such as the facial characteristics and palatopharyngeal disproportion, which may lead to VPI, have been assigned to the presence of platybasia, that is, an obtuse angulation of the cranial base.[2,4] Differences in the cranial base anatomy in individuals with VCFS, characterized by retrusion of the nasal and maxillary bones, were found on a cephalometric study.[5] However, the study did not find differences in the dimensions of the nasopharynx; thus, the authors suggested that VPI in these cases might be related to functional disorders. A magnetic resonance imaging study demonstrated that the thickness and diameter of the superior pharyngeal constrictor muscle are smaller in individuals with VCFS, constituting one of the main causes of pharyngeal hypotonia and consequently of the hypernasal speech observed in these individuals.[6]

Hypernasality is the most common speech disorder caused by VPI. Studies on individuals with VCFS report a frequency of 75% of hypernasality in these cases, most considered as severe, persistent, and difficult to evaluate, compared with individuals with cleft palate and/or VPI without VCFS.[7–14] However, even though hypernasality is an important manifestation of VPI, it is not necessarily related to the size of the velopharyngeal gap. When poor or absent velopharyngeal movement is observed, it is expected to find severe hypernasality. Conversely, there are several cases with small velopharyngeal gap and presence of severe hypernasality, which may be explained by the delayed time of velopharyngeal closure during speech in these cases, worsening the perception of the symptom.[15]

In addition to the hypernasality, the literature also indicates the high occurrence of compensatory articulation in individuals with

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VCFS, which, although not different than the articulation pattern observed in individuals with nonsyndromic cleft palate, are very different concerning the severity.[11,13,14] In addition, children with VCFS present poor articulation ability and even apraxic characteristics compared with children with DVF without the syndrome, which contributes to disturb the speech intelligibility.[14,16]

The complex manifestations of VPI in cases with VCFS complicate the establishment of the adequate therapeutic approach. Some surgical techniques indicated for treatment of severe VPI, such as the pharyngeal flap, should be carefully indicated in cases with VCFS due to the high prevalence (65%–85%) of abnormal positioning of the internal carotid arteries at the pharyngeal region, which coincides with the region where the pharyngeal pedicle is raised.[17,18] Diagnosis of this anatomic variation in these cases is fundamental; even though accomplishment of the pharyngeal flap is not impossible, other techniques should be considered in the presence of such alterations.[7,8,17–20]

The surgical techniques indicated for VPI correction in cases with VCFS include the techniques of Furlow[21] and Braithwaite and von Langenbeck with intravelar veloplasty.[22,23] These techniques use the procedure called intravelar veloplasty, whose main objective is the complete release of the palatal musculature, allowing replacement of the palatal vault muscles, which are posteriorly displaced to help in the velopharyngeal closure and are indicated when these muscles are inserted on the posterior border of the hard palate.[24] These techniques, either primary or secondary, regardless of the presence of VCFS, aim to modify the palatal anatomy to favor the velopharyngeal closure. Even when the velopharyngeal gap is not completely closed, the primary palatoplasty per se provides a better anatomic condition for the surgical success if a secondary surgical procedure is necessary.[25–27]

The literature has investigated the surgical outcomes of several techniques for VPI correction in VCFS. Tatum et al[17] described the speech and velar movement outcomes of 20 individuals with VCFS submitted to pharyngeal flap and observed elimination of hypernasality and complete velopharyngeal closure in 90% of cases. Similar outcomes were observed in another study, which reported 85% of balanced speech resonance or mild hypernasality in cases submitted to pharyngeal flap.[28] Discordantly, Widdershoven et al[12] used the technique of palatal lengthening and observed improvement in speech in only 42% of patients; according to the authors, no patients in this sample achieved normality. Other authors compared individuals with VCFS to individuals with nonsyndromic cleft submitted to pharyngeal flap and observed that the results in VCFS were worse compared with the group with nonsyndromic clefts.[9] The same authors also observed that, preoperatively, the hypernasality, nasal air escape, velopharyngeal gap, and nasalance were significantly more evident in cases with VCFS. A similar study demonstrated that surgery for VPI correction improved all aspects of speech analyzed, both in cases with VCFS and the others; however, the authors observed worse results for individuals with VCFS.[10]

In general, studies aiming to analyze the surgical outcome in cases with VCFS presented some limitations, including the retrospective design, small sample size, and poor definition of criteria of surgical success. Therefore, prospective studies analyzing the surgical outcome of VPI in cases with VCFS are required, to elucidate the factors that may interfere with the surgical outcomes and consequently allow the definition of the most adequate treatment for these cases.

This study investigated if individuals with VCFS present the same characteristics of speech and velopharyngeal function (VPF) compared with individuals with isolated submucous cleft palate and compared the effectiveness of surgery for VPI correction on the speech function and VPF, between the 2 groups.

METHODS

Subjects

Documentations of 191 patients with clinical signs of VCFS, registered from 1990 to 2009 in the Genetics and Speech Pathology Departments of Hospital for Rehabilitation of Craniofacial Anomalies/University of São Paulo (HRAC/USP) were analyzed. From this analysis, 25 individuals of both sexes, aged from 5 to 27 years, were selected, based on the following criteria: 6 VCFS clinical signs, at least no previous surgical treatment, and surgical indication for VPI. These patients composed the VCFS group as follows: 12 with submucous cleft palate, 8 with occult submucous cleft palate, and 5 presented congenital VPI. Six individuals were submitted to pharyngeal flap and 19 to palatoplasty with intravelar veloplasty.

For comparison purposes, a group of patients without syndrome with submucous cleft palate (SMCP) clinical signs of VCFS, presenting significant hypernasality and indication for primary palatoplasty, was evaluated. The SMCP group was selected from the hospital routine treatment at the same period of VCFS group. The individuals in both groups were matched for age and sex (Table 1).

Procedures

The patients were submitted to the following preoperative and postoperative procedures: digital recording of speech samples, nasometric evaluation, and nasoendoscopic evaluation of VPF. In the average, the evaluations were performed 6 days before and 17 months after surgery.

The speech samples were recorded in a digital system using a microphone (Superlux PRA-30; Superlux, Boulder, CO) connected to a microcomputer (Intel Pentium 4/256 RAM, soundcard Audigy 2–Sound Blaster, Creative; Intel, Santa Clara, CA), which were edited and later analyzed by the examiners. The hypernasality was scored by 3 examiners; all speech therapists are experienced with the evaluation of VPI. The hypernasality was scored using the 6-point scale,[29] in which 1 = absent, 2 = mild, 3 = mild/moderate, 4 = moderate, 5 = moderate/severe, and 6 = severe.

For the analysis, the 6-point scale was reduced to a 4-point scale, grouping the scores 3 and 4 in the moderate category (3) and scores 5 and 6 in the severe/intense category (4), thereby achieving the following scale: 1 = absent, 2 = mild, 3 = moderate, and 4 = severe.[30] In order to obtain a single value for each individual

TABLE 1. Sample Distribution According to Age and Sex in Each Group

		Age, y			Sex	
Groups	**n**	**Median**	**Minimum**	**Maximum**	**Female**	**Male**
VCFS	25	9	5	27	52% (n = 13)	48% (n = 12)
SMCP	25	9	4	24	52% (n = 13)	48% (n = 12)

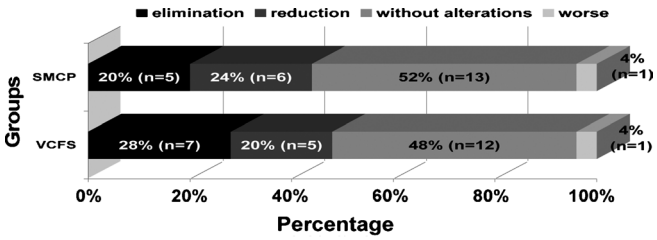

FIGURE 1. Distribution of patients according to postoperative hypernasality outcomes.

evaluated, the score assigned by most examiners was considered the final score of hypernasality.

The nasometric evaluation was performed using a nasometer model 6200-3 IBM (software version 30-02-3.22; Kay Elemetrics Corp), during reading of a sheet of sentences in Brazilian Portuguese, containing exclusively oral sounds.[31] The value of 27% was considered the upper limit of normality; that is, higher values were indicative of hypernasality.[32]

The nasoendoscopic evaluation of VPF was performed using a flexible endoscope (Olympus ENF-TYPE P3; Olympus. Tokyo, Japan). One plastic surgeon and 2 speech therapists experienced with this type of evaluation conducted the examination. The present study considered the results obtained during sentences with predominance of plosive and fricative sounds.[31] The VPF was analyzed considering the size of the velopharyngeal gap and classified according to the International Working Group Guidelines.[33] Thus, the dimension of the velopharyngeal gap was classified according to the movement of the palatal vault and pharyngeal walls during speech, using the following scores: 0 = absence of gap; 1 = minimum gap, with inconsistent contact of the velopharyngeal structures; 2 = small gap, that is, velopharyngeal space smaller than 50% in relation to the rest position; 3 = medium gap, velopharyngeal space around 50%; 4 = large gap, velopharyngeal space greater than 50%; and 5 = similar to the rest position. A single score was assigned to each individual in the preoperative and postoperative analyses. In case of variation of the gap size during the production of sentences, the larger gap was considered as the final score.

Data Analysis

Based on the score of hypernasality obtained on the evaluation by the examiners, the intraexaminer and interexaminer agreement was calculated using the κ coefficient.[34] The results obtained for each variable in the 2 groups were compared between the pre- and postoperative periods and between the 2 groups in both periods.

The success of surgery in the 2 groups was analyzed using an adaptation,[30] which considered for hypernasality and VPF: elimination, absence of hypernasality and velopharyngeal gap after surgery, i.e. reaching score one; reduction, when there was a reduction in the score in one or more points in relation to the preoperative period, yet not reaching score one; no alteration, when there was no change in the score between evaluations. Concerning the nasometry, the following criteria were considered: normalization, nasalance value ≤27% in the postoperative period; reduction, a reduction in the nasalance value higher than or equal to 8 percent points compared with the preoperative period, not reaching 27%; no alteration, when the value was equal or smaller than 8 percent points in relation to the preoperative period.

The comparison between percentages of improvement of variables between the preoperative and postoperative periods for both groups was performed by nonparametric Wilcoxon test, and the percentages of resolution were compared by the McNemar test.

The variables were also analyzed between the 2 groups in the postoperative period by the Kruskal-Wallis test. Comparison between the percentages of resolution in the same conditions was performed by the χ^2 test. A significance value of $P < 0.05$ was considered for all comparisons.

RESULTS

Hypernasality

In the preoperative and postoperative analysis of hypernasality of the 50 patients, the interexaminer agreement ranged from 0.55 to 0.67 (moderate to substantial), and the intraexaminer agreement varied from 0.75 to 0.92 (substantial to almost perfect).

According to the mean score obtained from analysis by the examiners before surgery, 20% of patients in the VCFS group presented mild hypernasality; 60%, moderate; and 20%, severe. In the SMCP group, 24% of the patients presented mild hypernasality; 56%, moderate; and 20%, severe. There was no statistically significant difference between groups at this period ($P = 0.690$).

After surgery, 28% of the VCFS group did not present hypernasality, 8% presented mild hypernasality, 56% presented moderate hypernasality, and 8% severe hypernasality. In the SMCP group, 20% did not present hypernasality, 32% presented mild hypernasality, 32% presented moderate hypernasality, and 16% presented severe hypernasality (Fig. 1). There was no statistically significant difference between groups in the postoperative period ($P = 0.232$).

Nasalance

A total of 39 patients were submitted to nasometric evaluation. Nine individuals in the VCFS group and 2 in the SMCP group did not perform this analysis because of poor compliance during examination.

Table 2 demonstrates the mean nasalance values obtained on the 2 periods, for the 2 groups. The mean nasalance before surgery in the VCFS group was 54% (SD, 8%), compared with 46% (SD, 11%) for the SMCP group, with no statistically significant difference between groups. After surgery, the mean nasalance in the VCFS group was reduced to 44% (SD, 15%), and the values in the SMCP group were reduced to 33% (SD, 15%). The mean nasalance values obtained after surgery were statistically smaller than the values obtained before surgery for the VCFS group ($P = 0.026$) and

TABLE 2. Mean (SD) Nasalance Scores, Obtained Before (Pre) and After (Post) Surgery in Both Groups

	Nasalance, %		
Groups	**Pre**	**Post**	**Pre × Post**
VCFS (n = 16)	54 (8)	44 (15)*	$P = 0.020$
SMCP (n = 23)	46 (11)	33 (15)*	$P = 0.000$

*Statistically significant difference between presurgery and postsurgery.

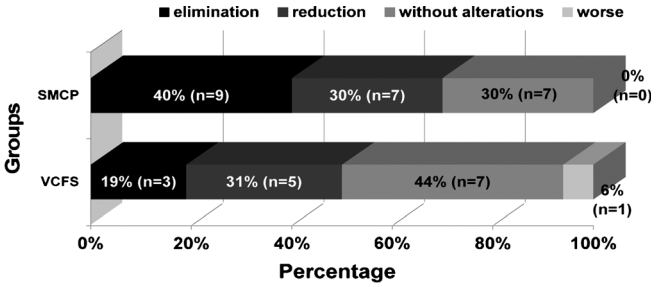

FIGURE 2. Distribution of patients according to postoperative nasalance scores outcomes.

for the SMCP group ($P = 0.000$). However, the comparison between postoperative values in the 2 groups demonstrated no statistically significant difference between groups.

Even though the mean nasalance values obtained in the 2 study groups did not reach normal values, the individual analysis of data demonstrated that 19% and 40% of patients in the VCFS and SMCP groups, respectively, presented normal nasalance after surgery. There was also reduction of nasalance in 31% and 30% of patients of the respective groups, without alterations in 44% and 30% of cases in the VCFS and SMCP groups, respectively (Fig. 2). However, the comparison between postoperative values in the 2 groups demonstrated no statistically significant difference between groups.

Velopharyngeal Function

Among the total 50 patients, 5 individuals in the SMCP group did not comply with the examination. Before surgery, 12% of patients in the VCFS group presented minimum velopharyngeal gap, 40% small, 12% medium, and 36% large. In the SMCP group, 50% of patients presented minimum velopharyngeal gap, 10% small, 15% medium, and 25% large, with no statistically significant difference between groups at this period ($P = 0.189$).

After surgery, it was observed that 8% of patients in the VCFS group presented complete velopharyngeal closure, and 44% exhibited minimum gap, 8% small, 8% medium, and 32% large. In the SMCP group, 25% of patients presented complete velopharyngeal closure, and 40% presented minimum gap, 5% small, 20% medium, and 10% large. The statistical analysis revealed statistically significant reduction in the size of the velopharyngeal gap in the VCFS and SMCP groups ($P = 0.019$ and $P = 0.049$), respectively. However, there was no statistically significant difference between groups concerning the size of the velopharyngeal gap in the postoperative period ($P = 0.131$).

Complete velopharyngeal closure (absence of velopharyngeal gap) was observed in 8% of patients in the VCFS group and 25% in the SMCP group. In 36% and 30%, there was reduction in the size of the velopharyngeal gap, respectively, for the same groups. In 52% and 40% of the respective groups, there was no alteration in the size of the velopharyngeal gap, as demonstrated in Figure 3. The statistical comparison between percentages of reduction, elimination and no alteration revealed no significant difference between groups concerning the modification of the velopharyngeal gap ($P = 0.505$).

DISCUSSION

A set of signs and symptoms characterizes a syndromic case, and it is often difficult to establish an accurate diagnosis without the participation of an interdisciplinary team, because of the variable expression of these signs and symptoms. The team should include clinical and laboratory genetic professionals; in the case of VCFS and several other syndromes, laboratory genetic examinations must be conducted to investigate the microdeletion in chromosome 22q11.

However, until completion of this study, the HRAC/USP did not have this resource; thus, molecular evaluation of individuals in the present sample was not possible. For this reason, the inclusion criteria comprised the presence of 6 or more clinical signs of VCFS; therefore, the expression "individuals with clinical signs of VCFS" was used to refer to these cases.

This study was proposed because of the need to verify if individuals with VCFS present the same characteristics of speech function and VPF compared with individuals with nonsyndromic isolated submucous cleft palate, as well as to compare the effectiveness of surgery for VPI correction on speech function and VPF between these 2 groups. Considering some particular characteristics in cases with VCFS, the therapeutic approach indicated for these cases has been a matter of concern in the daily routine of the hospital.

Several factors may affect the VPF in VCFS, including platybasia and absence or hypoplasia of pharyngeal tonsils, which increases the velopharyngeal space.[1,2] Associated with hypotonia and abnormality of the velopharyngeal muscles, this worsens the VPI; therefore, the pharyngeal flap technique is indicated in most cases.[12] Concomitantly, the risk of medial displacement of the internal carotid arteries poses risks for the accomplishment of this surgical technique in these individuals.[8,17]

Considering that speech is extremely important for social interaction and that the correct speech production requires anatomic and functional conditions of the velopharyngeal mechanism, analysis of this aspect in this population is fundamental. Therefore, the subjects in the current study were evaluated by auditory perceptual evaluation of speech, combined to instrumental evaluations of VPF using nasometry and nasoendoscopy.

With regard to the primary surgical treatment, the speech disorders persist in some individuals submitted to palatoplasty due to VPI, regardless of the presence of the syndrome. Depending on the severity, the symptoms impair the speech intelligibility, which may be even worse when associated to the inherent characteristics of VCFS, directly interfering with the quality of life of these patients.[35]

At HRAC/USP, most cases with nonsyndromic cleft palate are submitted to primary palatoplasty after 12 months of age. However, the primary surgery was performed later in the present patients, in the average at 9 years of age in both groups. The literature demonstrates that the late accomplishment of primary palatoplasty in cases with VCFS is common, considering that the diagnosis and onset of treatment occur between 4 and 17 years.[9,10,12,17,36] In individuals with SMCP without syndrome, especially not associated with overt cleft palate, the palatal alteration is often mild and may remain

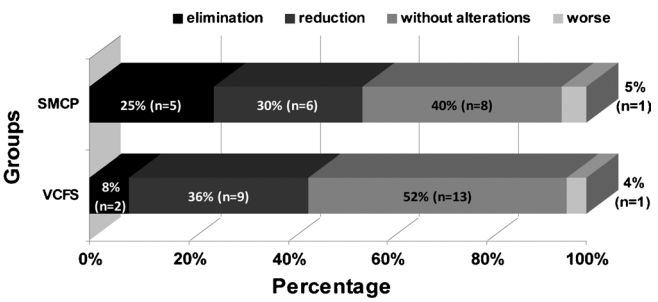

FIGURE 3. Distribution of patients according to postoperative velopharyngeal gap outcomes.

unnoticed until the family looks for treatment because of the manifestation of speech symptoms.

Therefore, the patients in this study, whose mean age on the first attendance at the institution was 8 years, initiated the treatment at a similar age as reported in the literature. It should be highlighted that most cases in this sample did not have a previous diagnosis or suspicion of VCFS before this attendance. It should be noticed that this age coincides with the onset of fundamental education, when the difficulties in interpersonal relationships and leaning are evidenced, which might remain unnoticed at the preschool age. However, in the clinical experience, when the family is questioned about the previous overall development, some difficulties are identified in several aspects including the speech, yet an expectant approach is often adopted.

A significant manifestation of speech of patients with VCFS is the alteration in speech resonance, such as the presence of hypernasality.[8–10,12,15,17,28,37] In the current study, alterations were observed in the function of the velopharyngeal mechanism preoperatively, which caused hypernasality, most frequently of moderate severity (60% and 56%) in the 2 study groups, which is of slightly higher proportions in the VCFS group, although not statistically significant.

The literature suggests that individuals with VCFS usually present greater VPI, yet this was not observed in the current study. In the preoperative period, there was no statistically significant difference between individuals in the VCFS and SMCP groups for the variables hypernasality and size of velopharyngeal gap. Different results were found concerning the nasal airflow values and size of velopharyngeal gap, observing significantly higher values in the preoperative period in cases with VCFS compared with patients without the syndrome.[9]

A possible explanation for the present results is the fact that the surgical indication for VPI correction was based on several evaluations, including the phenotypic clinical characteristics, auditory perceptive evaluation of speech, and instrumental evaluations, to define the adequate approach for each case. However, the patients presenting completely hypodynamic velopharyngeal mechanism were not selected for surgery, mostly being referred for placement of a palatal prosthesis, and thus were not included in this study. Therefore, the patients in this study, which were indicated for surgery, are those presenting the most favorable velopharyngeal conditions and thus are possibly similar to the cases of isolated cleft without the syndrome.

All individuals in this study were submitted to primary surgery for VPI correction, including intravelar veloplasty in 76% of patients in the VCFS group and 100% in the SMCP group. This procedure is a usual surgical procedure in primary palatoplasty management at HRAC/USP. Comparatively to pharyngeal flap, it is more physiological and is associated with low morbidity. Some authors consider that intravelar veloplasty must be the first option for the VPI treatment[21,38–40] due to improving muscular function. This is an important condition even for those patients who will require further intervention, such as pharyngeal flap, avoiding large flaps and its unfavorable effects.[26]

Literature has shown high success rates with intravelar veloplasty. Some authors has found 95% of VPI improvement,[38] whereas others reported similar proportions of 83% and 82%, respectively, after Furlow palatoplasty.[41,42] Reports in the literature suggest that intravelar veloplasty is more effective in patients with moderate VPI and small velopharyngeal gap,[39] a similar condition of most patients in the current study (52% in VCFS group and 60% in the SMCP group).

The surgical outcome in both study groups was analyzed according to the elimination or reduction of hypernasality and velopharyngeal gap, as well as the elimination of nasal air emission and compensatory articulations and normalization or reduction of the nasalance values. In general, the surgery promoted improvement/elimination of speech in most individuals in both groups. Therefore, individual analysis of the outcomes of each aspect of speech allows the evaluation of the positive effect promoted by surgery, which will be discussed later.

In the current study, the effect of surgery on hypernasality was positive, with reduction in nearly half of individuals in the VCFS (48%) and SMCP groups (44%), similar to the results of studies on patients with VCFS, which observed improvement in hypernasality in 42% of patients submitted to palatal lengthening.[12] Conversely, the results were worse than another study, which observed improvement in hypernasality in 66% of cases submitted to sphincteroplasty,[17] as well as compared with another study, which found improvement in 71% of cases submitted to the Furlow technique and sphincteroplasty.[10]

However, considering only the elimination of hypernasality as a positive result, it occurred in 28% of cases in the VCFS group and 20% in the SMCP group in the current study. These results are worse compared with studies on patients with VCFS that reported complete resolution of hypernasality in 90% of cases[17] and a study that reported resolution in 71%.[28] It should be highlighted that the percentages observed in both studies are related to individuals submitted to the pharyngeal flap surgery, whereas in the current study, only 6 patients in the VCFS group were submitted to this technique.

Based on the rationale that the pharyngeal flap provides better results, analysis of the 6 individuals in the VCFS group submitted to this technique in the current study revealed that 3 achieved oronasal balance of resonance, 2 presented reduction in hypernasality yet did not achieve normality, and only 1 maintained the same degree observed preoperatively. Another aspect to be considered is that the 3 patients who did not achieve normal values presented compensatory articulations, which directly interferes with the functionality of the velopharyngeal mechanism, as in any patient with VPI without the syndrome.

Even though hypernasality is often eliminated in patients with nonsyndromic cleft palate after surgical correction, some studies report worse speech outcomes in the population with VCFS.[9,11] Thus, an objective of this study was to analyze the effectiveness of surgery, comparing the results between the 2 groups.

Studies demonstrated worse degrees of hypernasality in patients with VCFS after surgery compared with individuals without the syndrome.[10,12] In the current study, the statistical analysis did not reveal significant difference between groups. An explanation for this outcome may be the fact that, among the 25 patients with VCFS analyzed, only 20% (5 patients) exhibited severe hypernasality in the preoperative period, which may have contributed to the lack of significance in the results of statistical analysis between groups.

Instrumental measurements were also used to analyze the VPF, including nasometry and nasoendoscopy, which are recommended to help and complement the perceptive evaluation.[33,43,44] Similar results were observed in nasometry in both study groups, with no statistically significant difference in preoperative and postoperative periods. These results disagree with a study that revealed higher nasalance values before surgery in individuals in the VCFS group, which the authors indicated as a cause for the worse surgical outcomes observed in these patients.[9]

Analysis of the nasalance results revealed that surgery provided a similar reduction in the VCFS and SMCP groups, but not achieving normality. The statistical analysis did not demonstrate significant difference between groups. This result corroborates the findings of a study on individuals with VCFS submitted to surgery for VPI correction, in which the authors observed statistically significant improvement in the nasalance values, yet not achieving normality.[12]

Another instrumental evaluation used in this study was nasoendoscopy of the VPF, which allows direct observation of the velopharyngeal mechanism from both anatomic and functional standpoints. Analysis of the size of velopharyngeal gap, measured on the nasoendoscopy, on the preoperative period did not reveal significant difference between groups, discordant from a previous study that reported greater velopharyngeal gap in children with VCFS on the preoperative period, with frequent need of secondary palatal surgery.[9] Analysis of the palatoplasty effect of the VPF revealed that, despite a reduction of the velopharyngeal gap in 44% in the VCFS group, the velopharyngeal gap was eliminated in only 8%, with no statistically significant difference concerning the size of the velopharyngeal gap between the 2 study periods. These results were worse than those obtained in a study that observed complete velopharyngeal closure in 71% of patients with VCFS submitted to surgical procedure on the palate and 29% who exhibited minimum velopharyngeal gap with mild hypernasality.[28]

In some cases, incompatibility was observed between the nasoendoscopy and the resonance scoring. In 5 patients in the VCFS group and 2 in the SMCP group, even though the resonance was scored as balanced, there was no elimination of the velopharyngeal gap. This type of incompatibility was discussed in a previous study that suggested that the velopharyngeal closure might occur at a lower place in the velopharyngeal region, impairing its complete observation.[45] Another explanation for this finding is that, in the presence of a small velopharyngeal gap, it may not be possible to detect alterations in resonance during the analysis of recordings, such as the mild hypernasality, which often remains unnoticed even by experienced examiners.[46,47]

Similar to the current study, discrepancies between the assessment of hypernasality and the findings of VPF were also reported.[12] The authors discussed that improvement of the velopharyngeal mechanism does not necessarily correspond to an improvement in speech, emphasizing the complexity of speech disorders observed in VCFS.

Concerning the postoperative outcomes of the VPF, the literature assigns the worse outcomes observed in individuals with VCFS to the deficient motor control of speech, with different potential in the time of velopharyngeal closure, which associated to other aspects as the structural abnormalities, may contribute to the speech alterations.[15] In the current study, elimination of the velopharyngeal gap was observed in only 8% of individuals in the VCFS group and 25% in the SMCP group, although with no statistically significant difference between groups. In a previous study, a significant difference in the velopharyngeal closure after surgery between patients with VCFS and patients without syndrome was observed.[12]

On the other hand, it should be considered that, although the complete resolution of VPI was observed in a small percentage of patients, significant improvement in VPF was observed in both groups with a velopharyngeal gap reduction in 36% of patients in the VCFS group and 30% in the SMCP group. It indicates that surgery improves VPF, creating favorable anatomic conditions for secondary intervention.

One limitation of the current study was the large age range of the sample, with patients in the early school period and adults, which may have influenced the outcomes. Another limitation refers to the time elapsed between surgery and the postoperative evaluations, which may have been insufficient, because the clinical experience has demonstrated that the effects of surgery for VPI correction are better observed after 18 months postoperatively. The literature reports that patients with VCFS require more time to correct their speech with the aid of speech therapy.[12] Therefore, these cases should be followed for longer periods, to observe their evolution with time.

Along more than 40 years of establishment of HRAC/USP, hundreds of patients with VCFS have been diagnosed at our institution. However, a small sample of this group could be included in the current study, especially because of medical problems inherent to the syndrome, which precluded the surgical VPI correction; the difficulties to achieve accurate speech samples in this population; and the lack of standardization of speech sample recording.

Therefore, one goal of this study was to evaluate if the surgical procedure for VPI correction is equally effective in individuals with VCFS. So far, reports in the literature on the surgical outcomes for the treatment of VPI on the speech of individuals with VCFS are relatively scarce, with some limitations, and usually without well-defined protocols. Based on the evaluations performed and considering the several alterations inherent to the syndrome, it was observed that the surgical procedure for VPI treatment in the VCFS provided improvement in several aspects of speech, even though only few cases achieved normalization of speech. In general, the individuals with VCFS were benefited from surgical treatment. The positive effect on the speech intelligibility in some cases, although minimum, may mean a large step in the rehabilitation for the patients and their families, an stimulus to continue searching for treatment, which raises the thought that the surgical approach should be considered and discussed with the team during the rehabilitation process, as well as the need to conduct larger studies on this population.

The need of a specialized team for the care of patients with VCFS is highlighted, especially in reference centers, including professionals to assist their multiple needs, taking part in the process and promoting the early diagnosis and informing the family in the prognosis and on the rehabilitation process, which extends up to adulthood in most cases. The rehabilitation of these patients aims to provide the maximum use of their abilities, minimizing their speech, learning, and emotional disorders, with a view to promote their social interaction, respecting their limitations.

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