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Comparison between two different approaches of surgical management for velopharyngeal insufficiency

Comparação entre duas abordagens diferentes de tratamento cirúrgico da insuficiência velofaríngea

Renata Paciello Yamashita¹, Camila Tomazi Rissato², Rafaeli Higa Scarmagnani³, Ana Paula Fukushiro^{1,4}, Inge Elly Kiemle Trindade^{1,5}

ABSTRACT

Introduction: Secondary palatoplasty with intravelar veloplasty (IV) may be performed before pharyngeal flap (PF) for surgical treatment of velopharyngeal insufficiency in individuals with cleft palate. This surgical approach aims to improve the velopharyngeal conditions, thus avoiding indication of a large pharyngeal flap and its undesirable effects on respiration. Purpose: To investigate the effects of pharyngeal flap performed after intravelar veloplasty for the treatment of velopharyngeal insufficiency on nasality and respiration. Methods: Analysis of postoperative outcomes of speech nasality and respiration in 50 individuals with repaired cleft palate and velopharyngeal insufficiency, being 23 submitted to pharyngeal flap after intravelar veloplasty (Group IV+PF) and 27 submitted only to pharyngeal flap (Group PF). Nasality was determined by nasometry, and the effect of surgery on respiration was assessed by measuring the minimum nasopharyngeal transverse section area, obtained by the pressure-flow technique and investigation of respiratory complaints, using a specific questionnaire. Statistical comparison between groups was performed by the Mann-Whitney test and Student t test, at a significance level of p<0.05. Results: There was no difference between groups for the outcomes of nasality, measurement of nasopharyngeal area and investigation of respiratory complaints. Conclusion: Both approaches for surgical treatment of velopharyngeal insufficiency demonstrated similar outcomes concerning the elimination of hypernasality, as well as for respiration, suggesting that accomplishment of pharyngeal flap after intravelar veloplasty did not provide better speech resonance outcomes nor favored respiration.

Keywords: Cleft palate; Velopharyngeal insufficiency; Speech; Speech disorders; Surgical procedures, Operative

RESUMO

Introdução: A palatoplastia secundária com veloplastia intravelar (VI) pode ser realizada previamente ao retalho faríngeo (RF), para o tratamento cirúrgico da insuficiência velofaríngea em pacientes com fissura palatina. Esta abordagem cirúrgica tem como finalidade melhorar as condições velofaríngeas, evitando, assim, a indicação de um retalho faríngeo largo e seus efeitos indesejáveis sobre a respiração. Objetivo: Investigar os efeitos do retalho faríngeo realizado após a veloplastia intravelar para tratamento da insuficiência velofaríngea sobre a nasalidade e a respiração. Métodos: Análise dos resultados pós-cirúrgicos da nasalidade de fala e da respiração de 50 indivíduos com fissura de palato reparada e insuficiência velofaríngea, sendo 23 submetidos ao retalho faríngeo após a veloplastia intravelar (Grupo VI+RF) e 27 submetidos unicamente ao retalho faríngeo (Grupo RF). A nasalidade foi determinada pela nasometria e o efeito da cirurgia sobre a respiração foi analisado pela medida da área de secção transversa mínima nasofaríngea, obtida pela técnica fluxo-pressão e pelo levantamento das queixas respiratórias, utilizando questionário específico. A comparação estatística entre os grupos foi realizada por meio do teste Mann-Whitney e teste t de Student, considerando significante o valor de p<0,05. **Resultados:** Verificou-se que não houve diferença entre os grupos para os resultados de nasalidade, da medida da área nasofaríngea e do levantamento de queixas respiratórias. Conclusão: Ambas as abordagens para tratamento cirúrgico da insuficiência velofaríngea mostraram resultados equivalentes, no que se refere à eliminação da hipernasalidade, bem como à condição da respiração, sugerindo que a realização do retalho faríngeo após a veloplastia intravelar não levou a melhores resultados de ressonância de fala e não favoreceu a respiração.

Palavras-chave: Fissura palatina; Insuficiência velofaríngea; Fala; Distúrbios da fala; Procedimentos cirúrgicos operatórios

 $Study\ conducted\ at\ the\ Laboratory\ of\ Physiology\ of\ the\ Hospital\ for\ Rehabilitation\ of\ Craniofacial\ Anomalies,\ Universidade\ de\ S\~ao\ Paulo\ -USP\ -Bauru\ (SP),\ Brazil.$

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Conflict of interest: No

Authors' contribution: *RPY* main author, in charge of the study project, study design and general supervision of the stages of accomplishment and manuscript preparation; *CTR* in charge of the study, data collection and analysis and manuscript writing; *RHS* collaboration in data collection and manuscript writing; *APF* collaboration in data analysis and manuscript writing; *IEKT* collaboration in manuscript writing and review.

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INTRODUCTION

The adequate velopharyngeal functioning depends on the anatomical integrity and synchronized movement of soft palate and lateral and posterior pharyngeal walls, and is fundamental for normal speech production. When the velopharyngeal function is altered, part of the air flow is deviated to the nasal cavity, leading to the appearance of symptoms that may impair the speech in different manners. The failure in velopharyngeal closure caused by structural disorders is called velopharyngeal insufficiency (VPI)^(1,2).

VPI leads to speech disorders, such as hypernasality, nasal air emission, weak intraoral pressure and compensatory articulation. Hypernasality is characterized by the excess nasal resonance in the production of non-nasal sounds, caused by communication between the oral and nasal cavities, and is considered the most representative symptom of VPI⁽²⁾. VPI is primarily diagnosed by auditory perceptual speech analysis, which is considered the main indicator of clinical significance of VPI symptoms and, as such, is fundamental in the diagnosis of velopharyngeal insufficiency^(3,4). However, since this is a subjective method, it should be complemented by instrumental evaluation, either direct or indirect, which allows determining the cause, extent and location of velopharyngeal dysfunction⁽⁴⁾. Correction of the speech symptoms of VPI requires secondary palatal surgery in most cases^(5,6,7).

Several surgical techniques have been described for that purpose, the most common of which are the achievement of pharyngeal flaps and sphincteroplasty, which are well reported in the literature, and secondary palatoplasty with intravelar veloplasty, which has been used more recently for correction of VPI^(7,8,9,10,11). In general, selection of the surgical technique is based on the structural and functional conditions of the velopharynx determined on preoperative evaluation, including the extent and mobility of the palatal vault, movement of pharyngeal walls and type of velopharyngeal closure^(7,12).

The pharyngeal flap surgery comprises creation of a myomucous tissue bridge between the posterior pharyngeal wall and the soft palate, determining two lateral orifices. The objective of surgery is to create a mechanical obstruction to the passage of air between the oropharynx and nasopharynx, maintaining two lateral orifices to allow efficient nasal respiration at rest. During speech, the orifices should close by the action of lateral pharyngeal walls, to avoid nasal air escape and hypernasality, thereby directing the airflow to the oral cavity^(7,9,12). There is consensus in the literature about the high success rate of pharyngeal flap surgery to reduce or eliminate the speech symptoms caused by VPI^(12,13,14). However, there is concern among clinicians and investigators in this field about surgeries that change the anatomy of the velopharyngeal region and alter the nasopharyngeal permeability, such as the pharyngeal flap^(9,13,15). For this reason, the literature has advocated surgical techniques that allow adequate velopharyngeal closure in a condition more similar to the normal anatomical conditions, i.e. without altering the anatomy of the velopharyngeal sphincter, thus reducing the morbidity risks^(8,16,17,18). One procedure used for that purpose is intravelar veloplasty^(7,19,20,21). The main objective of this surgical procedure is to reposition the soft palate musculature as posteriorly as possible, thus providing good mobility to the palatal vault and consequently improving the velopharyngeal competence^(7,22). For that purpose, extended dissection of the soft palate muscles is performed, completely detaching them from the bony edge of the hard palate, separated from the nasal and oral mucosa and united in the midline, as a muscle sling, in more posterior positioning. Since this is a muscular repositioning procedure, intravelar veloplasty may be incorporated to conventional techniques already used for secondary palatoplasty. The main criterion to be considered to select individuals for intravelar veloplasty should be the anteriorized attachment of the palatal musculature and presence of small velopharyngeal gap^(7,17,22). Several studies demonstrated good speech outcomes in individuals with VPI submitted to intravelar veloplasty^(8,16,17,18,20,21).

It is currently established in the literature that pharyngeal flap is indicated for individuals with severe VPI, i.e. presenting large gaps in velopharyngeal closure. Conversely, intravelar veloplasty is more effective in cases of less severe VPI, individuals with small velopharyngeal gaps and/or marginal velopharyngeal function. However, intravelar veloplasty has been also shown as effective in the presence of severe VPI(17,20,21). Even though intravelar veloplasty does not completely solve the speech symptoms in individuals with large gaps, some authors advocate the utilization of this procedure as first option for the treatment of VPI, since this is considered a more physiological procedure. Therefore, individuals requiring pharyngeal flap at a later moment, after veloplasty, may present a more favorable velopharyngeal condition, thus avoiding the indication of a broad flap and its undesirable effects on respiration(17,23).

Therefore, this study compared the results of pharyngeal flap surgery performed after palatoplasty with intravelar veloplasty (two-stage surgical approach) with the outcomes of pharyngeal flap surgery routinely performed at the hospital (single surgical stage), on speech nasality and respiration.

METHODS

Sample selection

The study was approved by the Institutional Review Board of the Hospital for Rehabilitation of Craniofacial Anomalies, *Universidade de São Paulo* (HRAC-USP) (report n. 734.759). The study analyzed the outcomes of speech nasalance and respiration in 50 individuals with repaired cleft palate, with or without cleft lip, submitted to correction of VPI, being 23 individuals initially submitted to intravelar veloplasty and

then to pharyngeal flap after 34 months in the average (Group IV+PF). The time elapsed between intravelar veloplasty and pharyngeal flap ranged from six months to three years, for most individuals. For only three individuals this period was five, seven and nine years, respectively. Group PF was composed of 27 individuals submitted only to pharyngeal flap. The age of individuals ranged from 11 to 43 years (mean age 25 years) in group IV+PF, and between 7 and 40 years (mean age 22 years) in group PF. Concerning the type of cleft in both groups, ten individuals presented isolated cleft palate, eight unilateral cleft lip and palate and five exhibited bilateral cleft lip and palate, in group IV+PF. In group PF, ten individuals presented isolated cleft palate, ten unilateral cleft lip and palate and seven bilateral cleft lip and palate. All individuals (in both groups) presented VPI diagnosed by auditory perceptual speech analysis and nasoendoscopy, thus they had indication for surgical correction of VPI. Before pharyngeal flap surgery, all individuals exhibited nasalance values indicating hypernasality, and nasopharyngeal section area values indicative of normality. With regard to the respiratory symptoms, analysis of postoperative outcomes in both groups considered only symptoms initiating after surgery (appearance of symptoms), or worsening of preexisting symptoms, observed with greater frequency or intensity, according to the reports of individuals or their caretakers.

Procedures

The study analyzed postoperative outcomes related to speech nasality, determined by the measurement of nasalance, by nasometry, and the outcomes related to respiration, assessed by measurement of the minimum nasopharyngeal transverse section area (nasopharyngeal area) by the pressure-flow technique, and investigation of respiratory symptoms of individuals using a specific questionnaire.

Measurement of nasalance – nasometry

The nasalance (acoustic correlate of nasality) was measured by nasometry. The system is composed of two microphones, located on each side of a sound separation plate positioned on the individual's upper lip. A helmet keeps the assembly in place. During reading of a standard text, the upper microphone captures signs of the nasal component of speech, while the lower microphone captures signs of oral component, which are filtered, digitized and analyzed on a specific software. Nasalance is calculated as the numeric ratio between nasal acoustic energy and total acoustic energy (sum of nasal and oral acoustic energy), multiplied by 100⁽⁵⁾. The examination is performed while reading a sequence of five sentences in Portuguese, containing exclusively oral sounds, for identification of hypernasality: Papai caiu da escada. Fábio pegou o gelo. O palhaço chutou a bola. Tereza fez pastel. A árvore dá frutos e flores. Individuals who are unable to read the text are asked to repeat each sentence, after the examiner. A value of 27% is considered as the upper limit or normality, i.e. values higher than 27% are considered as indicative of hypernasality⁽⁵⁾. Figure 1 presents a schematic drawing of the system setting.

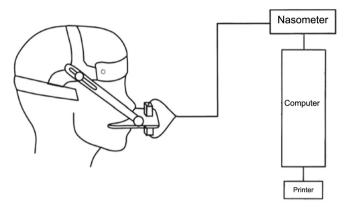


Figure 1. Scheme illustrating the instrumentation for measurement of nasalance (Nasometer 6200-3 IBM, Kay Elemetrics Corp.®, Lincoln Park, NJ, USA)

Source: Trindade IEK, Yamashita RP, Bento Gonçalves CGA. Diagnóstico instrumental da disfunção velofaríngea. In: Trindade e Silva Filho. Fissuras labiopalatinas: uma abordagem interdisciplinar. São Paulo: Editora Santos; 2007. p. 134.

Measurement of nasopharyngeal area – pressureflow technique

The minimum nasopharyngeal transverse section area (nasopharyngeal area) was obtained by the pressure-flow technique. This technique is based on the principle that a constriction area may be estimated by simultaneous measurement of the differential pressure between the two sides of constriction and the airflow crossing it (5,24). The nasopharyngeal area is measured during respiration at rest, positioning a catheter inside the oral cavity and another inside the nostril with smaller nasal flow, held in place by an obturator. Both catheters measure static air pressures, which are transmitted to pressure transducers. The nasal airflow is measured by a plastic tube, adapted to the nostril with greater flow, connected to a heated pneumotachograph and also connected to a pressure transducer (Figure 2).

The signals of the three transducers (nasal pressure, oral pressure and nasal flow) are transmitted to the computed system PERCISARS, version 4.01 (Microtronics Corp.®), for analysis on a specific software. The measurements are obtained on peaks of inspiration and expiration flow, in two to four successive respirations. The area considered for analysis corresponds to the mean of these multiple measurements and is calculated by the equation: A=V/k ($2\Delta P/d$)1/2, in which A=0 orifice area in cm²; V=0 nasal flow in cm³/s; V=00.65; V=01.047cm² was considered as the limit or normality V=01.047cm² was considered as the limit or normality V=02.

Investigation of respiratory symptoms – specific questionnaire

The respiratory symptoms reported by the individuals

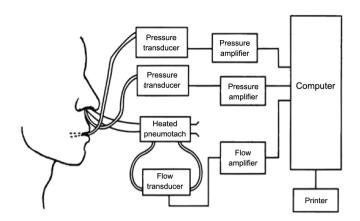


Figure 2. Instrumentation to determine the velopharyngeal orifice area (PERCI-SAR system, Microtronics Corp.®, Chapel Hill, NC, USA)

Source: Trindade IEK, Yamashita RP, Bento Gonçalves CGA. Diagnóstico instrumental da disfunção velofaríngea. In: Trindade e Silva Filho. Fissuras labiopalatinas: uma abordagem interdisciplinar. São Paulo: Editora Santos; 2007. p. 137.

or their caretakers were investigated using a standardized questionnaire specific for respiratory signs and symptoms. This study considered as respiratory symptoms only reports of appearance or worsening (increased frequency or intensity) of symptoms, such as oral respiration, snoring and sleep respiratory disorder, after surgery⁽⁹⁾.

Data analysis

The postoperative nasalance values between the two groups were statistically compared by the Mann-Whitney test. The nasopharyngeal area values and total number of respiratory complaints reported were compared between groups by the Student t test. All analyses were performed at a significance level of 5% (p<0.05).

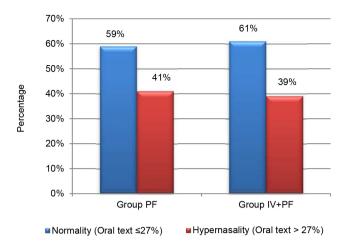
RESULTS

Comparisons of postoperative outcomes between groups PF and IV+PF

Speech nasality – measurement of nasalance

The mean nasalance value obtained in group PF after surgery was 27±12.4%, indicative of normality (8%-52%), compared to 24±15.27% in group IV+PF, also indicative of normality (6%-58%). In group PF, 59% (16) of individuals presented nasalance scores indicating normality (<27%) and 41% (11) exhibited scores indicating hypernasality. In group IV+PF, 61% (14) of individuals presented normal nasalance, while 39% (9) of individuals exhibited nasalance scores indicating hypernasality (Figure 3).

Comparison between the mean nasalance values between groups PF and IV+PF revealed no difference between the two groups (p=0.502), as demonstrated in Table 1.



Subtitle: Group PF = group pharyngeal flap; Group IV+PF = group intravelar veloplasty + pharyngeal flap

Figure 3. Percentage of individuals distributed according to the nasalance scores achieved after surgery in the group pharyngeal flap and group intravelar veloplasty + pharyngeal flap

Table 1. Statistical analysis of mean values of postoperative nasalance: comparison between groups pharyngeal flap and intravelar veloplasty + pharyngeal flap

Nasalance						
Group	Mean	SD	p-value			
PF (n=27)	27	12.4	0.512			
IV+PF (n=23)	24	15.27				

Mann-Whitney test (p<0.05)

Subtitle: n = number of individuals; Group PF = group pharyngeal flap; Group IV+PF = group intravelar veloplasty + pharyngeal flap; SD = standard deviation

Respiration – Measurement of nasopharyngeal area

The mean nasopharyngeal area of group PF was 0.636±0.274 cm² (0.115 cm²-1.200 cm²) after surgery, and 0.649±0.181 cm² (0.233 cm²-1.153 cm²) do for group IV+PF. Both groups exhibited reduced nasopharyngeal area values compared to the normative value, yet compatible with the presence of pharyngeal flap.

Comparison of the mean values of nasopharyngeal area between groups PF and IV+PF revealed no difference between groups (p=0.886) (Table 2).

Table 2. Statistical analysis of mean postoperative values of nasopharyngeal area: comparison between groups pharyngeal flap and intravelar veloplasty + pharyngeal flap

Nasopharyngeal area						
Group	Mean	SD	p-value			
IV+PF	0.649	0.181	0.886			
PF	0.636	0.274				

Student t test (p<0.05)

Subtitle: Group PF = group pharyngeal flap; Group IV+PF = group intravelar veloplasty + pharyngeal flap; SD = standard deviation

Investigation of respiratory symptoms

Isolated analysis of the three symptoms according to the individuals' reports revealed, in group PF, occurrence of 59% (16) of oral respiration, 63% (17) of snoring during sleep and 11% (3) of sleep respiratory disorder. In group IV+PF, oral respiration was observed in 61% (14), snoring in 69% (16) and sleep respiratory disorder in 9% (2).

Analysis of data revealed no difference between groups (p=0.503) concerning the occurrence of respiratory complaints after surgery, as presented in Table 3.

Table 3. Statistical analysis of the total number of reported respiratory complaints: comparison between groups intravelar veloplasty + pharyngeal flap and pharyngeal flap

Respiratory complaints							
Group	OR	S	SRD	Total	p-value		
IV+PF	14	16	2	32	0.503		
PF	16	17	3	36			

Student t test (p<0.05)

Subtitle: Group PF = group pharyngeal flap; Group IV+PF = group intravelar veloplasty + pharyngeal flap; OR = oral respiration; S = snoring; SRD = sleep respiratory disorder

DISCUSSION

The effects of pharyngeal flap and secondary palatoplasty with intravelar veloplasty on the correction of speech symptoms(12,17,18,20,21) secondary to VPI and on the respiration of individuals (9,23,25) have been widely investigated and reported in the literature, in isolated manner. One of the few studies comparing the outcomes of these two surgeries on speech⁽¹⁰⁾ demonstrated that pharyngeal flap was more efficient than intravelar veloplasty for reduction of hypernasality and adequacy of velopharyngeal closure in individuals with residual VPI. Conversely, pharyngeal flap changes the anatomy of the velopharyngeal region and thus alters the permeability of the nasopharynx, possibly leading to undesirable respiratory symptoms as oral respiration, snoring, obstructive sleep respiratory disorders, and even hyponasality^(9,15,23,26,27). In all these studies, surgeries were performed as the single procedure for correction of VPI. As previously demonstrated⁽⁹⁾, among individuals assisted at the Hospital for Rehabilitation of Craniofacial Anomalies, Universidade de São Paulo (HRAC-USP), 36% of individuals submitted to pharyngeal flap began to present respiratory symptoms, such as oral respiration, snoring and feeling of respiratory difficulty during sleep, due to the reduction of nasopharyngeal dimensions after surgery. A systematic review of the main studies in the literature investigating the obstructive potential of the pharyngeal flap revealed high ratio (84%) of middle-aged adults presenting signs and symptoms of obstructive sleep apnea⁽²⁶⁾. Recently, a polysomnography study demonstrated that a significant number of middle-aged

adults with repaired cleft presented obstructive sleep apnea and related respiratory symptoms, even in the absence of pharyngeal flap⁽²⁷⁾.

No previous studies have investigated the effect of surgical treatment for correction of VPI, as analyzed in the present study, i.e. the effect of pharyngeal flap performed after intravelar veloplasty, which was the scope of the present study. The objective was to compare, by instrumental evaluation, the postoperative outcomes of speech resonance (nasality) and respiration between a group of individuals with VPI submitted to pharyngeal flap, months after accomplishment of intravelar veloplasty, and a group submitted only to pharyngeal flap surgery. In these cases, veloplasty was performed to improve the velopharyngeal conditions and avoid the indication of a broad flap, since all individuals exhibited severe VPI, i.e. presented large velopharyngeal gap, as determined by consensus among the team and diagnosed by preoperative nasoendoscopy.

The speech nasality was analyzed by nasometry results. This method is recommended for the diagnosis and follow-up of velopharyngeal insufficiency^(12,13,20,28). The method comprises measurement of nasalance, a physical measurement that corresponds to the relative quantity of acoustic energy emitted by the nasal cavity during speech. This is a non-invasive technique, easy to perform and useful for evaluation of resonance, since it complements the perceptual evaluation and the observations of direct evaluation methods.

However, data interpretation should consider that nasality and nasalance indicate different phenomena and that perception of nasality is influenced by several factors that do not influence the acoustic measurement⁽⁵⁾. However, the efficiency of nasometry to correctly identify deviations in nasality detected by auditory perceptual speech analysis was assessed by retrospective analysis of data collected from 194 individuals with repaired cleft lip and/or palate⁽⁵⁾. This study revealed that the sensitivity and specificity of nasalance scores to correctly identify individuals with and without hypernasality was 0.78 and 0.79, respectively. That is to say, 78% of individuals identified with hypernasality, by perceptual analysis, presented high nasalance scores (sensitivity), and 79% of individuals without deviation in nasality in the perceptual analysis exhibited reduced nasalance scores (specificity). Thus, a high score was defined as a value higher than 27%. The overall efficiency of the method was 0.78, indicating that 78% of individuals were scored equally by nasometry and by subjective evaluation⁽⁵⁾.

Nasometry revealed that, in the average, both study groups exhibited nasalance scores within the limit of normality (\leq 27%) after surgery. The ratio of individuals without nasality in group IV+PF was slightly higher than in group PF. However, this difference was not significant, demonstrating that the accomplishment of pharyngeal flap after intravelar veloplasty does not assure better speech resonance results.

The respiration of individuals after surgery was analyzed from the measurements of nasopharyngeal area, obtained by the pressure-flow technique. Considering that this method reflects the anatomical and functional changes promoted by surgery and provides objective information on how the flap influences nasal permeability, this is fundamental to evaluate the effects of this type of surgical intervention (9,12,28).

The results demonstrated that both groups exhibited mean values of nasopharyngeal area below the limit of normality, which is expected after pharyngeal flap surgery. Also, there was no significant difference between the study groups. Individual analysis of data demonstrated that 87% (20) of individuals in group IV+PF and 85% (22) in group PF presented values of nasopharyngeal area below the limit of normality, which suggests that the accomplishment of pharyngeal flap, after intravelar veloplasty, does not assure the achievement of a narrower flap that may reduce the risks of mechanical nasopharyngeal obstruction.

The effects of both surgeries on the respiration of individuals were also analyzed from complaints concerning respiratory symptoms that appeared or worsened after surgery. This study specifically analyzed the reports of individuals regarding the appearance or worsening of frequent oral respiration, snoring and sleep respiratory disorder. The specific questionnaire applied revealed 59% and 61% individuals with oral respiration, 63% and 69% with snoring and 11% and 9% reporting sleep respiratory disorder, for groups PF and IV+PF, respectively. There was no difference between groups concerning the appearance or worsening of respiratory complaints after surgery.

The present results demonstrated that both surgical approaches using the pharyngeal flap presented similar results concerning hypernasality and respiration. In an ultimate analysis, surgical correction of VPI performed in two stages (secondary palatoplasty with intravelar veloplasty, and pharyngeal flap after some months) did not provide better results in the elimination of hypernasality, nor demonstrated lower ratio of individuals with respiratory complaints after surgery, compared to pharyngeal flap as single surgical approach. These findings suggest that, for individuals presenting severe VPI, as confirmed by preoperative perceptual and instrumental evaluation, accomplishment of pharyngeal flap as single surgical approach should be considered for correction of speech symptoms secondary to VPI.

CONCLUSION

Both approaches for surgical treatment of velopharyngeal insufficiency exhibited similar outcomes concerning the elimination of hypernasality, as well as for respiration, suggesting that accomplishment of pharyngeal flap after intravelar veloplasty did not provide better speech nasality outcomes, nor better conditions for respiration.

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