




Could the speech resonance of individuals with cleft lip and palate be affected by complete section of pharyngeal flap?

A secção cirúrgica do retalho faríngeo pode comprometer a ressonância de fala de indivíduos com fissura lábio palatina?

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ABSTRACT

Purpose: To investigate the effect of complete section of pharyngeal flap on speech hypernasality. **Methods:** The study analyzed twenty-six individuals with repaired cleft palate±lip underwent pharyngeal flap surgery to treat velopharyngeal insufficiency and posteriorly underwent complete section of the flap due to the occurrence of respiratory symptoms. Hypernasality was determined by auditory-perceptual speech assessments and nasometry at 18 months after surgery, on average. Hypernasality was perceptually classified as: 1 = absent or 2 = present and determined by nasalance measurement during reading of sentences containing exclusively oral sounds, considering a cutoff of 27% ($p \leq 0.05$). **Results:** Perceptual assessment before section of the flap revealed that eight (31%) individuals showed normal resonance, while 18 (69%) presented hypernasality. After surgery, one (4%) subject remained with normal resonance and 25 (96%) presented hypernasality. According to nasometry, before surgery, 13 (57%) individuals presented nasalance scores lower than 27%, indicative of absence of hypernasality (mean = $15 \pm 8\%$) and ten (43%) presented nasalance scores indicative of hypernasality (mean = $41 \pm 7\%$). After surgery, four (17%) patients remained with scores indicative of absence of hypernasality (mean = $19 \pm 10\%$) and for 19 (83%) the nasalance scores were indicative of hypernasality (mean = $45 \pm 7\%$). There was no difference between perceptual and nasometric speech evaluations. **Conclusion:** Surgery for complete section of pharyngeal flap caused deterioration of speech resonance, leading to the reappearance of hypernasality in most patients in this study.

Keywords: Cleft palate; Velopharyngeal insufficiency; Speech; Speech perception; Speech disorders

RESUMO

Objetivo: Investigar o efeito da cirurgia para secção completa do retalho faríngeo sobre a hipernasalidade de fala. **Métodos:** Foram avaliados 26 indivíduos com fissura de palato±lábio reparada, submetidos à cirurgia de retalho faríngeo para tratamento da insuficiência velofaríngea e que, em função do aparecimento de queixas respiratórias, necessitaram nova cirurgia para secção do retalho. A hipernasalidade foi determinada por meio das avaliações perceptiva e nasométrica da fala 18 meses, em média, após a secção do retalho. Na avaliação perceptiva, a hipernasalidade foi classificada como: 1 = ausente ou 2 = presente e, na nasometria, foi determinada por meio da medida da nasalância durante a leitura de sentenças contendo, exclusivamente, sons orais, considerando-se, como limite de normalidade, o escore de 27% ($p \leq 0,05$). **Resultados:** A avaliação perceptiva mostrou que, antes da secção do retalho, oito (31%) indivíduos apresentavam ressonância equilibrada e 18 (69%) apresentavam hipernasalidade. Após a cirurgia, um (4%) paciente permaneceu com ressonância equilibrada e 25 (96%) apresentaram hipernasalidade. De acordo com a nasometria, antes da cirurgia, 13 (57%) indivíduos apresentaram valores de nasalância inferiores a 27%, indicando ausência de hipernasalidade (média = $15 \pm 8\%$) e dez (43%) pacientes apresentaram valores indicativos de hipernasalidade (média = $41 \pm 7\%$). Após a cirurgia, quatro (17%) pacientes permaneceram com valores indicativos de ausência de hipernasalidade (média = $19 \pm 10\%$) e 19 (83%) apresentaram valores de nasalância indicativos de hipernasalidade (média = $45 \pm 7\%$). Diferença entre as avaliações perceptiva e nasométrica da fala não foi observada. **Conclusão:** A cirurgia para secção completa do retalho faríngeo causou deterioração da ressonância de fala, levando ao reaparecimento da hipernasalidade, na maioria dos pacientes estudados.

Palavras-chave: Fissura palatina; Insuficiência velofaríngea; Fala; Percepção da fala, Distúrbios da fala

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INTRODUCTION

Individuals with cleft palate may present symptoms that impair the speech intelligibility, even after primary surgery for palate repair. These symptoms are caused by velopharyngeal dysfunction (VFD), being hypernasality the most characteristic symptom⁽¹⁻³⁾. The treatment of VFD for elimination of speech symptoms requires secondary palate surgery. The several techniques described in the literature for that purpose include the pharyngeal flap with superior pedicle (PF)⁽⁴⁻⁸⁾. This technique involves the connection between soft palate and posterior pharyngeal wall by construction of a myomucous tissue bridge, with base at the level of the adenoid tissue (superior pedicle), a region considered with greatest medial movement of the lateral pharyngeal wall in most cases. The PF is delimited by two lateral orifices, whose diameter and permeability allow nasal breathing at rest and act as sphincters during speech^(5,9,10). Thus, the mechanical obstruction created between the oropharynx and nasopharynx reduces the air passage to the nasal cavity during production of oral sounds, leading to elimination or at least reduction of speech symptoms caused by VFD⁽⁵⁻⁸⁾.

Despite the benefits for speech, in some cases, the PF is associated with obstruction of the upper airway, due to reduction of nasopharyngeal dimensions, possibly causing respiratory symptoms as mouth breathing, snoring and obstructive sleep apnea^(6,8,11,12). One study conducted at this service, aiming to analyze the effects of PF on breathing in the long term, evidenced, by aerodynamic assessment, significant reduction of nasopharyngeal dimensions and presence of persistent respiratory complaints (present more than one year after surgery) in 36% of individuals with cleft palate⁽¹¹⁾. More recently, another study investigated the frequency and severity of obstructive sleep apnea by polysomnography examination, comparing adults following PF surgery and adults without the flap. The authors reported that the proportion of individuals with respiratory symptoms was higher in the group with PF (77%) compared to patients without flap (60%), even though there was no significant difference between the study groups⁽¹²⁾.

Also aiming to investigate the harmful effects of PF, yet related to speech, investigators in the same service analyzed the occurrence of hyponasality secondary to obstructive PF. The authors evaluated 159 individuals with cleft palate to investigate the influence of PF surgery on the production of nasal sounds and observed appearance of hyponasality in 14% and 25% of patients after surgery, according to perceptual assessment and nasometry, respectively⁽¹³⁾.

In cases of obstructive PF in which the respiratory complications do not respond to treatment, such as the use of continuous positive air pressure machine (CPAP)⁽¹⁴⁾, or revision surgery of PF with widening of lateral orifices, a surgery for complete section of the pharyngeal flap may be necessary⁽¹⁴⁻¹⁶⁾.

The effect of PF on the speech^(7,13,17,18) and respiration^(8,11,12,17,18) have been widely analyzed. However, little has been investigated about the harmful effects of flap section on the speech resonance.

In one of the first reports on the speech outcomes⁽¹⁹⁾, the authors evaluated, by perceptual assessment, nasoendoscopy and videofluoroscopy, nine children without hypernasality, following surgery for total or partial section of pharyngeal flap, due to respiratory complications. The authors observed that two among the nine children presented return of the symptom. The other seven patients remained with normal resonance. According to

the authors, the fact that resonance remains unaltered in most cases may be explained by factors as adherence of flap to the posterior pharyngeal wall, or persistence of broad flap even after partial section, as demonstrated by instrumental examinations.

Other investigators used the flow-pressure technique as an instrument to determine the velopharyngeal closure in 12 individuals, before and after surgery for section of pharyngeal flap. The authors observed that, before surgery, all participants presented adequate velopharyngeal closure, indicating absence of hypernasality, and after surgery 80% (8) of individuals remained with adequate closure. Two patients (20%) began to present inadequate velopharyngeal closure, according to the velopharyngeal area values. The authors considered that the good result, even after flap removal, may have occurred due to anatomical changes in the nasopharyngeal region, which persisted even after removal of the pharyngeal flap⁽¹⁵⁾.

Considering the lack of studies analyzing the effects of pharyngeal flap section on speech, this study investigated the effect of surgery for complete section of the pharyngeal flap, performed due to the appearance of respiratory complaints after correction of velopharyngeal insufficiency, on the speech resonance. In the last analysis, it aimed to verify if section of the flap worsened the resonance, as identified by the appearance of hypernasality.

METHODS

Retrospective study, conducted at the Laboratory of Physiology at the Hospital for Rehabilitation of Craniofacial Anomalies at University of Sao Paulo – HRAC-USP, Bauru (SP), Brazil, after approval by the Institutional Review Board (protocol n. 74/98).

Sample

The study analyzed data from 26 patients with repaired cleft palate±lip and PF (15 operated in the institution where the study was conducted and nine in another service), of both genders, aged 12 to 51 years (mean 36±13 years), underwent surgery for complete section of pharyngeal flap due to respiratory difficulties reported after pharyngoplasty. The age at PF surgery for individuals operated in the institution ranged from 5 to 49 years (mean 30±18 years). The surgical procedure for flap section was performed between three to 11 years (mean 7±3 years) after its accomplishment. For individuals whose PF had been performed in another institution, there was no information about the date of the procedure, thus it was not possible to establish the time elapsed between PF and its section.

The study included individuals with PF following section of myomucous flap, due to significant respiratory complaints that appeared after PF surgery, such as snoring, respiratory arrest during sleep and shortness of breath. The study did not include individuals with syndromes and/or other craniofacial anomalies associated with cleft lip and palate, or clinical evidences of nasal obstruction upon examination that might impair their outcome.

Procedures

The study analyzed the results of perceptual and nasometric assessments of speech, performed before PF section (PRE) and on average 18±16 months (six months to six years) after this procedure (POST), according to the routine of attendance of the institution. The postoperative evaluations are ideally performed six to 12 months after the surgical procedure, which is considered sufficient for complete healing of the posterior pharyngeal wall, where the pharyngeal flap pedicle was sectioned. Among the individuals analyzed, 15 returned to outpatient follow-up on the established period and nine returned after the maximum period established for outpatient follow-up at the institution.

Perceptual assessment da hypernasality

The hypernasality was scored by presential evaluation, by a single examiner, as: 1 = absent hypernasality (normal oronasal resonance), 2 = present hypernasality.

Nasometric evaluation of speech: nasometry

The nasalance (acoustic correlate of nasality) was determined using a nasometer model 6200-3 IBM (software version 30-02-3.22), composed of two microphones separated by a metallic plate, which catch the signs of nasal and oral components of speech. The examination is performed while reading five sentences containing exclusively oral sounds of Brazilian Portuguese. As the text, presented on the screen of a microcomputer connected to the system, is read, the signals are caught by the microphones and nasalance is calculated by the numeric ratio between the nasal acoustic energy and the total acoustic energy (sum of nasal and oral acoustic energies), multiplied by 100. The value of 27% is considered the cutoff value for normality. Thus, values higher than 27% are considered indicative of hypernasality. Figure 1 schematically shows the system settings.

Data analysis

Hypernasality was expressed as scores and nasalance as percentages. The scores of hypernasality before and after section of PF were compared by the Wilcoxon test. The mean nasalance values, in the two situations, were compared by the Student t test for paired samples. Comparison between the two types of evaluation was performed by the McNemar test. All tests considered a significance level of $p < 0.05$.

RESULTS

According to the perceptual assessment of speech, in the situation PRE, 31% (8/26) of patients presented normal resonance, while 69% (18/26) presented hypernasality. In the situation POST, 4% (1/26) remained with normal resonance and 96% (25/26) presented hypernasality. The individual analysis of data demonstrated that 7 individuals began to present hypernasality after section of PF. Statistical comparison

demonstrated difference between the percentages of individuals in conditions PRE and POST ($p < 0.001$). The ratio of patients presenting normal resonance and hypernasality before and after section of PF is shown in Table 1.

The nasometry data were analyzed for a total of 23 patients, since 3 individuals did not perform this evaluation after surgery. According to the nasometric evaluation, in the situation PRE, 57% (13/23) of patients presented nasalance values indicative of normality (absence of hypernasality), with mean nasalance of 15±8% and 43% (10/23), indicating hypernasality (mean nasalance 41±7%). In the situation POST, 17% (4/23) of patients remained with values indicative of normality (mean = 19±10%) and 83% (19/23) presented hypernasality (mean = 45±7%). This indicates that, according to nasometry, 9 individuals exhibited worsening of speech resonance, with appearance of hypernasality after section of PF. Statistical analysis revealed that, after section of pharyngeal flap, the patients presented significantly greater mean nasalance values, compared to those obtained before surgery ($p < 0.001$), as demonstrated in Table 2.

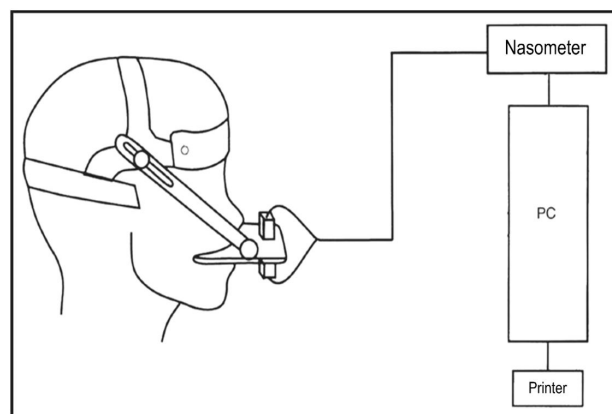


Figure 1. Scheme representing the instrumentation for nasalance measurement (Nasometer 6200-3 IBM, Kay Elemetrics Corp., Lincoln Park, NJ, USA)⁽²⁰⁾

Table 1. Distribution of percentage (number) of patients according to the presence and absence of hypernasality determined by perceptual assessment, before and after surgery for section of pharyngeal flap

HYPERNASALITY (Perceptual Assessment)		
Condition (n)	Absent	Present
PRE (26)	31% (8)	60% (18) [#]
POST (26)	4% (1)	96% (25) [#]

[#]Before vs after: $p < 0.001$ - Wilcoxon test

Subtitle: PRE = before section of pharyngeal flap; POST = after section of pharyngeal flap

Table 2. Distribution of percentage (number) of patients according to the presence and absence of hypernasality, determined by nasometry before and after surgery for section of pharyngeal flap

HYPERNASALITY (Nasalance)		
Condition (n)	Absent	Present
PRE (23)	57% (13)	43% (10) [#]
POST (23)	17% (4)	83% (19) [#]

[#]Before vs after: $p < 0.001$ - Student t test

Subtitle: PRE = before section of pharyngeal flap; POST = after section of pharyngeal flap

Comparison between the two types of evaluation, perceptual and nasometric assessments of speech, concerning the proportion of individuals with and without hypernasality after pharyngeal flap section, demonstrated no significant difference between the results for both ($p = 1.000$).

DISCUSSION

Among the surgical techniques described in the literature for the treatment of velopharyngeal dysfunction, pharyngeal flap surgery with superior pedicle still presents high success rates concerning the elimination of speech symptoms^(7,21-24).

Since this technique alters the anatomy of the velopharyngeal region, creating an obstruction between oropharynx and nasopharynx, the pharyngeal flap may lead to the appearance of respiratory symptoms that are often permanent⁽¹¹⁾. When the respiratory symptoms affect the quality of life of individuals, surgery for full section of the pharyngeal flap is necessary.

This study analyzed the effect of surgery for pharyngeal flap section on speech resonance, to investigate if removal of the pharyngeal flap worsened the resonance. That is to say, the study aimed to investigate whether individuals with pharyngeal flap and normal resonance began to present hypernasality as a consequence of flap section. For that purpose, the study used two assessment methods well known in the literature: perceptual assessment of speech and nasometry.

Though subjective, perceptual assessment is considered the gold standard for evaluation of speech in individuals with cleft palate and the main indicator of clinical significance of this symptom^(1,25,26). One limitation of the present study is that the presence or absence of hypernasality was defined based on presential evaluation performed by a single examiner during routine follow-up of these patients, whose data were registered. It is known that, in such studies, perceptual assessment should ideally be performed by consensus among different examiners, using audio or video recordings of speech samples, which was not available for individuals analyzed in the present study. However, it should be noted that all perceptual assessments were performed by professionals (examiners) of the institution, with extensive experience in cleft lip and palate.

Among the instrumental methods, nasometry is one of the most correlated with perceptual assessment of nasality^(3,27) and thus is part of routine exams performed to follow the treatment of individuals with cleft palate.

Although there was little variation between the results of nasometry and perceptual assessment, statistical comparison showed no difference between them, confirming the good correlation between the two assessment modalities. However, since this examination analyzes even slight deviations of resonance⁽²⁸⁾, the presence of occasional nasal congestion resulting from acute or chronic allergic respiratory processes, unidentified during examination, might lead to unreliable results of nasometric evaluation. Therefore, although hypernasality is perceptible to the human ear, upper airway obstruction, even without obvious symptoms, precludes the instrument from capturing, even partially, the necessary nasal acoustic energy to calculate the nasalance, which may indicate absence of speech symptom.

Although it was not a goal of the present study, analysis of the records of respiratory symptoms reported by the patients during routine care identified that three among the four

individuals who remained with normal nasalance values after removal of the flap occasionally reported nasal obstruction, mouth breathing, and allergic processes. Thus, the difference in the percentage of individuals with normal resonance after surgery, verified among the evaluation methods, may be partly explained by the presence of respiratory symptoms not detected during evaluation of these patients.

Data analysis showed that, according to the perceptual assessment of nasality, among eight patients with normal resonance before section of the pharyngeal flap, only one remained with unaltered resonance. The remaining seven presented hypernasality, i.e. removal of the pharyngeal flap returned the patients to the VFD condition.

Regarding the results of nasometry, the mean nasalance values obtained before section of the pharyngeal flap were 26%, indicating that, on average, the individuals analyzed had normal nasalance value, although this value is considered very close to the cutoff limit of normality (27%). However, individual data analysis revealed that, among the 23 individuals, 13 had normal values ranging from 5% to 14%. Among these, only four remained in the same condition, i.e. without speech symptom after section of the pharyngeal flap. For the other nine individuals, the nasalance values became indicative of hypernasality after flap removal. The mean nasalance value, after surgery, became 40%, thus indicative of hypernasality. The significant increase in mean nasalance after surgery demonstrates that removal of the pharyngeal flap led to the onset of hypernasality in most patients.

There is broad literature regarding the success of pharyngeal flap surgery to eliminate the symptoms of velopharyngeal dysfunction^(18,23,29) and concerning the appearance of respiratory difficulties resulting from this surgical technique^(8,11,12,15). Other studies have also advocated surgery to increase the lateral orifices of the flap, in an attempt to minimize or eliminate respiratory problems and, sometimes, speech problems, such as hyponasality^(16,30). However, this study analyzed the speech results in cases in which the respiratory problems in this population were managed by section of the pharyngeal flap, an issue scarcely addressed in the literature.

Though scarce, the literature demonstrates satisfactory results after section of the pharyngeal flap, concerning the speech resonance^(14,15). However, the present study demonstrated, by perceptual and instrumental assessment, that for most patients, removal of the flap, which provided normal speech resonance, led to the reappearance of velopharyngeal functioning patterns prior to surgery, causing the appearance of hypernasality. However, in a small part of patients (4% according to perceptual assessment and 17% according to nasometry), removal of the flap did not alter the resonance.

It is assumed that changes in anatomical and functional conditions achieved by the flap persisted after their removal, somehow favoring the velopharyngeal closure. This may be a result of fibrous scar tissue, formed by removal of the flap that remains in the posterior pharyngeal wall, functioning as a support (enlargement of the posterior wall) and aiding the velopharyngeal closure⁽¹⁴⁾. Another explanation for this result is that, while the flap remained in place, there may have been "learning" of the velopharyngeal mechanism to achieve complete velopharyngeal closure and such behavior was maintained even after flap removal. It may be assumed that this "learning" effect was more evident in those individuals who remained with the pharyngeal flap for longer time. However, individual analysis

of data revealed that the individual who remained with the pharyngeal flap for longer time (11 years) returned to present hypernasality after flap section, while the only individual who remained with normal resonance after section of the pharyngeal flap was precisely the one who remained with the myomucous tissue bridge for shorter time (three years) until its complete section. This evidences that a possible “learning” of the velopharyngeal mechanism may not occur in all cases following flap section. Additionally, the time during which the individual remains with the pharyngeal flap does not seem to be a determining factor for the occurrence of such “learning”. Future studies should be conducted to further evaluate these findings.

It should be highlighted that all individuals were submitted to evaluation after the section of the flap by the multidisciplinary team of the institution, to define the most appropriate management for the treatment of persistent speech symptoms or resulting from the removal of the pharyngeal flap. These approaches include surgical procedures, such as a new pharyngeal flap or palatoplasty with intravelar veloplasty; fitting of palatal prosthesis (pharyngeal bulb) and speech therapy for elimination of compensatory articulations.

CONCLUSION

The perceptual and nasometric assessments of speech demonstrated that surgery for complete section of pharyngeal flap worsened the velopharyngeal function, leading to the appearance of hypernasality in most individuals analyzed. However, though in a small part of individuals, sectioning the flap did not cause disorders in speech resonance. This finding may be explained by several factors, such as persistent anatomical changes, formation of fibrous cicatricial tissue that may contribute to the velopharyngeal closure during speech, or even an effect of learning of velopharyngeal performance.

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