

# **Original Article** Artigo Original

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# Keywords

Cleft Palate Orthognathic Surgery Velopharyngeal Insufficiency Speech Speech Disorders

Surgical maxillary advancement and speech resonance: comparison among cleft types

Avanço cirúrgico de maxila e ressonância de fala: comparação entre os tipos de fissura

# ABSTRACT

Purpose: This study investigated the influence of the cleft type on the appearance of hypernasality after surgical maxillary advancement (MA). Methods: Nasality was determined by measurement of nasalance (acoustic correlate of nasality) by nasometry. The study involved analysis of the nasalance scores of 17 individuals with isolated cleft palate (CP), 118 with unilateral cleft lip and palate (UCLP) and 69 with bilateral cleft lip and palate (BCLP), of both sexes, aged 18 to 28 years, after MA. Only individuals with normal nasalance scores indicating balanced resonance before MA were included in this study. Nasometry was performed 3 days before and 15 months after MA, on average. The proportion of patients who presented nasalance scores indicating hypernasality after surgery was calculated by the ANOVA test, and comparison among the different cleft types was evaluated by the chi-square test ( $p \le 0.05$ ). **Results:** No significant difference was found in the proportions of individuals with hypernasality among the cleft types. Conclusion: Nasometry showed that the appearance of hypernasality after MA in individuals with cleft palate with or without cleft lip occurred in similar proportions, regardless of the cleft type.

## **Descritores**

Fissura Palatina Cirurgia Ortognática Insuficiência Velofaríngea Fala Distúrbios da Fala

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## **RESUMO**

Objetivo: Investigar a influência do tipo de fissura sobre o aparecimento da hipernasalidade após o avanço cirúrgico da maxila (AM). Método: A nasalidade foi determinada por meio da medida de nasalância (correlato acústico da nasalidade) utilizando-se a nasometria. Foi realizada a análise dos escores de nasalância de 17 indivíduos com fissura isolada de palato (FP), 118 com fissura de lábio e palato unilateral (FLPU) e 69 com fissura de lábio e palato bilateral (FLPB), de ambos os sexos, com idades entre 18 e 28 anos, submetidos ao AM. Apenas indivíduos com escores de nasalância indicativos de ressonância equilibrada previamente ao AM foram incluídos neste estudo. A nasometria foi realizada, em média, três dias antes e 15 meses após o AM. A proporção de pacientes que apresentaram escores de nasalância indicativos de hipernasalidade após o AM foi calculada por meio do teste ANOVA e a comparação entre os diferentes tipos de fissura foi realizada utilizando-se o teste Qui-quadrado (p < 0.05). **Resultados:** Não foi observada diferença significante entre as proporções de indivíduos com hipernasalidade, de acordo com o tipo de fissura. Conclusão: A nasometria mostrou que o aparecimento da hipernasalidade após o AM, em indivíduos com fissura de palato envolvendo ou não o lábio, ocorreu em proporções similares independentemente do tipo de fissura.

Study conducted at Laboratório de Fisiologia, Hospital de Reabilitação de Anomalias Craniofaciais - HRAC, Universidade de São Paulo - USP - Bauru (SP), Brasil.

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## INTRODUCTION

The synergistic action of the velar and pharyngeal walls muscles promote adequate velopharyngeal closure (VPC), allowing balanced oronasal resonance during speech. In the presence of structural alterations, as in the case of cleft lip and palate (CLP), the VPC may only be possible after accomplishment of the palatal repair surgery<sup>(1)</sup>. The influence of factors as age, surgical technique and type of cleft on the velopharyngeal function after primary palatoplasty is well established in the literature<sup>(2,3)</sup>.

Concerning the impact of cleft type, studies using the classification system of *Veau* investigated the relevance of the cleft type on the velopharyngeal function after primary palatoplasty and observed that complete VPC was present in a greater number of patients with unilateral cleft lip and palate (UCLP) and incomplete cleft palate (ICP) compared to those with bilateral cleft lip and palate (BCLP) and complete cleft palate (CCP). The authors suggested that the type of cleft may affect the VPC due to the influence of the vomer on the velar length. The insertion of velar muscles on the vomer in UCLP and ICP favors the muscle tissues at the velopharyngeal region, thus allowing complete VPC, when compared to the palatal velar muscles in BCLP, in which the vomer is fully isolated, without any muscle attachment. Also, in this type of cleft, the bony palate is structurally shorter, which may directly affect the length and mobility of the velum<sup>(4,5)</sup>.

Also, the iatrogenic potential of the rehabilitation process is not similar to the different types of clefts. Factors as congenital dysmorphology of the midface, functional adaptation, iatrogenic effects of surgery, and bony structural variations among the different types of CLP may cause an important influence on midface growth, leading to maxillomandibular discrepancies (MMDs). Thus, midface growth may occur in a different manner, depending on the cleft type and severity<sup>(6-8)</sup>. This occurs mainly due to the scar fibrosis caused by the several surgical procedures to which individuals with CLP are submitted during childhood, which may affect the midface growth<sup>(9)</sup>. Studies have demonstrated that MMDs occur more frequently in individuals with complete cleft lip and palate compared to patients with isolated cleft palate<sup>(2,10)</sup>.

In case of moderate to severe MMDs, it is necessary to perform surgical treatment by repositioning of the bone bases, to enhance the orofacial functions  $^{(2,10,11)}$ , which occurs in nearly 10 to 50% of the population with CLP<sup>(9,12)</sup>. The type of surgery most frequently performed for correction of MMDs is the Le Fort I maxillary osteotomy for maxillary advancement (MA), which may be combined to osteotomies of the mandible and chin<sup>(13,14)</sup>. Studies emphasized that MA, by reestablishing the maxillomandibular balance and consequently adjusting the positioning of tongue and teeth, promotes a beneficial effect for speech production<sup>(15,16)</sup>. However, even though it has been confirmed that the extent of maxillary advancement, in millimeters, does not directly influence the speech results in individuals with CLP(17-19), the forward movement of hard and soft palate achieved during MA surgery may change the pattern of VCP, contributing to the appearance of hypernasality as a consequence of dysfunction of the velopharyngeal mechanism (VPD)<sup>(11, 20)</sup>.

The diagnosis of hypernasality is mainly achieved by auditory perceptual evaluation of speech, considered the "gold standard" to evaluate the symptoms related to VPD. However, due to its subjective nature the auditory perceptual evaluation is often associated with an instrumental evaluation to define the approach to be followed  $^{(1,21,22)}$ .

Nasometry is an objective and quantitative method for speech analysis that presents good relationship with the perceptual assessment of nasality<sup>(1,21,23)</sup>. This technique allows indirect determination of speech resonance by the measurement of nasalance (acoustic correlate of nasality). This measurement, expressed in percentage, is determined by measurement of the acoustic energy simultaneously captured from the oral and nasal cavities during speech. The values obtained by this examination suggest the presence or absence of hypernasality and its strong correlation with the auditory perceptual assessment has been demonstrated in previous studies<sup>(1,7,21,23,24)</sup>.

Considering the different anatomical conditions that involve the palate, healing processes after primary surgeries for palatal repair and their impact on the growth and development of soft and hard structures, the present study hypothesis was that speech impairment, after this surgical procedure, are proportional to the extent and severity of the cleft. That is, individuals affected by BCLP would be more susceptible to the appearance of hypernasality after MA. Thus, this study investigated the influence of cleft types involving the palate on the appearance of speech hypernasality after MA, using nasometry as an objective and quantitative method for speech analysis.

#### **METHODS**

This study was conducted at the Laboratory of Physiology of the Hospital for Rehabilitation of Craniofacial Anomalies, University of São Paulo – HRAC – USP, Bauru (SP), Brazil, after approval by the Institutional Review Board (IRB) (n. 291.650). The study was retrospective and analyzed data obtained from the patient records, thus a signed informed consent form was not necessary according to the IRB.

#### Casuistic

The study analyzed data from 204 individuals with repaired cleft palate with or without cleft lip, submitted to surgical maxillary advancement, being 17 with isolated cleft palate (CP), 118 with UCLP and 69 with BCLP, of both sexes, aged 18 to 28 years ( $22.9\pm4.1$ ). The study included only patients presenting MMD with anterior and/or posterior crossbite (overjet smaller than or equal to 0mm) and scores indicating absence of hypernasality (balanced speech resonance) obtained by nasometry before surgery. Patients with syndromes and/or other craniofacial anomalies related to CLP, or even submitted to secondary palatal surgery for correction of velopharyngeal dysfunction before MA were not included in this study.

#### Procedures

The study analyzed nasalance scores obtained on average three days before (PRE-MA) and 15 months (12 months to 2 years and 9 months) after surgery (POST-MA), following the routine of care of the institution. The postoperative evaluations are ideally performed 12 months after surgery, which is considered enough time for complete bone healing and adaptation of the velopharyngeal musculature.

#### Nasometry

The nasalance scores were obtained for all individuals using a nasometer (Model 6200-3 IBM, version 30-02-3.22; Kay Elemetrics) while reading a sequence of five sentences containing exclusively oral sounds of Brazilian Portuguese, aiming to identify the hypernasality: "Papai caiu da escada. Fabio pegou o gelo. O palhaço chutou a bola. Tereza fez pastel. A árvore dá frutos e flores"<sup>(22)</sup>. The score of 27% wads considered to indicate the limit of normality<sup>(24)</sup>. Thus, values higher than 27% suggested the presence of hypernasality.

#### Statistical analysis

Nasalance was expressed as percentage. Differences among groups were calculated by the ANOVA test. The proportion of patients presenting hypernasality after surgery was compared among cleft types by the chi-square test. The study considered a significance level of p < 0.05.

# RESULTS

According to nasometry, 79% (162/204) of patients had nasalance values indicating absence of hypernasality after MA, while 21% (42/204) of all patients presented nasalance scores indicating hypernasality. From the total of 42 patients with appearance of hypernasality, 18% (3/17) had CP, 17% (20/118) UCLP and 27% (19/69) BCLP, as shown in Table 1.

Comparison among the proportion of patients presenting hypernasality after surgery, for each cleft type, did not reveal significant difference among groups, as demonstrated in Table 2.

**Table 1.** Number of individuals according to cleft type and presenceor absence of hypernasality after surgical maxillary advancement andmean nasalance values (±SD) for each group

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	HYPERNASALITY AFTER MA			
Cleft type	N (≤27%)	N (>27%)	Mean Nasalance (%)	ANOVA test
СР	14	3	21±11%	
UCLP	98	20	21±12%	0.0927
BCLP	50	19	21±12%	
Total	162	42	21±12%	

Caption: SD = Standard Deviation; MA = Surgical maxillary advancement; CP = isolated cleft palate; UCLP = unilateral cleft lip and palate; BCLP = bilateral cleft lip and palate; N ( $\leq$  27%) = number of patients with nasalance scored indicating absence of hypernasality; N (> 27%) = number of patients with nasalance scored indicating hypernasality. Non-significant difference among groups – ANOVA test (p = 0.955)

 Table 2. Comparison among the proportions of patients presenting

 hypernasality after surgical maxillary advancement according to cleft type

	HYPERNASALITY AFTER MA			
		Chi-square test	p-value	
CP vs UCLP	18% vs 17%	0.075	0.785	
CP vs BCLP	18% vs 27%	0.277	0.598	
UCLP vs BCLP	17% vs 27%	2.350	0.125	

Caption: MA = Surgical maxillary advancement; CP vs UCLP = isolated cleft palate vs unilateral cleft lip and palate; CP vs BCLP = isolated cleft palate vs bilateral cleft lip and palate; UCLP vs BCLP = unilateral cleft lip and palate vs bilateral cleft lip and palate. Non-significant difference among groups after surgery – chi-square test

#### DISCUSSION

The surgical maxillary advancement is a procedure employed to establish adequate orofacial functions by repositioning of bone bases. However, in individuals with cleft lip and palate, worsening of the velopharyngeal function has been reported after this surgical procedure. Several studies investigated the effect of MA on the speech of individuals with repaired cleft palate, using perceptual<sup>(13-15,17-19)</sup> and instrumental<sup>(11,17,18)</sup> evaluations for that purpose.

Investigators in the field conducted a critical literature review and analyzed 39 papers published between 1971 and 2004, which investigated the effects of craniomaxillofacial osteotomies and distraction osteogenesis on the speech and velopharyngeal function. Among these, 32 studies were related to MA. The authors found 12 papers that concluded that MA does not present clear harmful effects for the speech and velopharyngeal function. Other 15 studies evidenced report of speech resonance impairment due to the surgical procedure, and in nine of these 15 studies these damages were observed in a small part of individuals analyzed. The other five studies reported worsening of velopharyngeal function only in individuals with borderline VPC<sup>(25)</sup>.

It is known that, in isolation, the extent of maxillary advancement in millimeters does not directly influence the speech outcomes in individuals with CLP<sup>(17-20)</sup>; however, the literature is controversial concerning the possible causes that lead to worsening of velopharyngeal function after Le Fort I osteotomy<sup>(17)</sup>. Aiming to investigate the morphofunctional aspects of the velopharyngeal region that may be considered predictors of the appearance or worsening of hypernasality after surgical maxillary advancement, a recent study investigated the speech outcomes of 52 patients with repaired CLP submitted to Le Fort I osteotomy. For that purpose, perceptual assessment of speech was performed combined to subjective and tomographic evaluation of the morphofunctional aspects of the palate. The authors observed that 21% (11/52) of individuals analyzed started to present speech symptoms, and this result was correlated to regular or poor mobility of the velum. That is, the worse the velum movement to complete the VPC, the worse is the speech outcome after MA<sup>(19)</sup>.

Despite this unprecedent result, it is known that other variables may be related to the risk of worsening of velopharyngeal function after orthognathic surgery and must be investigated. Among the variables previously analyzed in the literature for that purpose, no study investigated the influence of cleft type on the speech outcomes after this surgical procedure.

Ultimately, the aim of this study was to investigate if the appearance of hypernasality after surgical maxillary advancement occurs in different proportions depending on the type of cleft (CP, UCLP, BCLP) of the individual. For that purpose, speech resonance was analyzed by nasometry, an instrument that accurately reflect the results of perceptual auditory assessment of hypernasality<sup>(1,21,23,24)</sup>.

The results demonstrated that 21% (42/204) of all patients presented hypernasality after surgical maxillary advancement. This percentage is similar to the speech results reported by other authors. Chanchareonsook et al.<sup>(17)</sup> observed that two out of eight

(25%) patients submitted to surgical maxillary advancement presented nasalance scores indicating hypernasality after surgery. Haapanen et al.<sup>(26)</sup> demonstrated that 27% of 15 individuals with CLP submitted to Le Fort I osteotomy for the treatment of Class III MMD presented worsening of velopharyngeal function, which was detected by perceptual auditory assessment of speech. However, this result is lower than 45% of hypernasality observed by Trindade et al.(11) in 22 individuals submitted to MA combined to procedures involving the mandible and nose. However, it should be considered that these speech results were observed in an average period of nine months after surgery. The literature has demonstrated that surgical stability is achieved, at least, one year after maxillary advancement<sup>(27)</sup>. The accommodation of soft tissues at the velopharyngeal region may occur slowly and take a long period to stabilize, leading to a late response concerning the final outcome of speech resonance<sup>(14)</sup>. This difference in the proportion of individuals presenting hypernasality after surgery may be explained by the period of postoperative evaluation, which was 15 months in the average in this study. Other investigators demonstrated that, in a group of 10 individuals submitted to MA, none presented changes in speech resonance after the surgical procedure, according to the nasalance values. However, only four patients in the study group presented balanced resonance before surgery<sup>(18)</sup>, which should be considered in the interpretation of results observed.

Concerning the proportion of individuals with hypernasality after surgery, no significant difference was observed among the three groups. Despite the statistical results, analysis of individual data of nasalance demonstrated that the appearance of hypernasality was slightly higher in the group of patients with BCLP (27%) compared to groups with UCLP (17%) and CP (18%). The literature has indicated some particularities concerning the velopharyngeal region of individuals with BCLP.

Recently, a study using magnetic resonance images analyzed the nasopharyngeal volume and linear measurement of nasopharyngeal depth, length and thickness of the velum in three groups of children with different cleft types. The authors observed that individuals with BCLP demonstrated greater nasopharyngeal volume compared to those with UCLP and submucous cleft. No difference was observed among groups concerning the linear measurements<sup>(28)</sup>.

Similarly, other study compared changes in the pharyngeal region after maxillary osteotomy in 50 patients with UCLP, BCLP and CP using cephalometric analysis and observed significant change in the nasopharyngeal area in the three groups after surgery. Even though most measurements were greater in the group of patients with CP before and after surgery, the most evident change in the nasopharyngeal region was observed in the group with BCLP (88%) compared to individuals with UCLP (83%) and CP (73%)<sup>(7)</sup>.

Considering that changes in the nasopharynx tend to be more evident in individuals with BCLP after MA, as demonstrated by Heliövaara et al.<sup>(7)</sup>, it is possible to consider the hypothesis that the shape of the nasopharyngeal region or even the behavior of muscles that limit this region present peculiar characteristics that may lead to different speech results. This might explain the tendency that a greater number of individuals in the group with BCLP presented hypernasality after MA compared to the other groups.

Based on the assumption that individuals with UCLP and CP may present different muscular conditions and favorable concerning the size, position and physiology, compared to those with BCLP<sup>(4)</sup>, it is reasonable to assume that the latter group may be more vulnerable to speech damage after MA. However, this was not demonstrated in the present study. It is believed that, in individuals presenting the speech symptom after MA, the functional conditions of the palate may be inadequate as a consequence of primary palatoplasty, and may indicate the population that presents regular or poor mobility of the velum, which has been demonstrated to lead to the appearance or worsening of hypernasality after MA, regardless of the cleft type involving the palate<sup>(19)</sup>. Other explanation for this result is that the appearance of hypernasality may be related to differences in the shape of the velopharyngeal region. It may be assumed that individuals in which this region is narrower present more favorable conditions for VPC after MA compared to those with wider shape of the velopharyngeal region, which may negatively influence the velopharyngeal function.

It should be highlighted that the present study observed a significant proportion of individuals (21%) presenting hypernasality after surgery. This group deserves special attention in the planning of future surgical interventions aiming to correct residual speech symptoms.

It should also be mentioned that the surgical techniques employed at the moment of primary palatoplasty were not controlled and were performed by more than one plastic surgeon. Similarly, the maxillary advancement surgeries were performed by more than one maxillofacial surgeon, and the surgical techniques employed were not controlled, i.e. advancement of two or more segments, total maxillary advancement or even bimaxillary surgery. Considering that the type of primary palate surgery, surgeon's experience and type of procedure performed for forward maxillary movement may influence the surgical results concerning the accommodation of soft tissues after orthognathic surgery, this was considered a limitation of the present study. However, all plastic surgeons and maxillofacial surgeons in the institutional team present minimum experience of 9 years in the accomplishment of palate repair surgeries and orthognathic surgeries.

Other limitation of the study was the non-inclusion of perceptual auditory assessment of speech in the study methodology, especially because this is the "gold standard" for the evaluation of VPD symptoms. This occurred because the perceptual auditory assessments of speech of these individuals were performed in person by a single examiner. Considering that, for research purposes, it is advisable to achieve the results of subjective evaluations by consensus between two or more examiners, the authors of the present study decided to perform instrumental evaluation as a method of analysis of results.

Overall, the present study demonstrated that the appearance of hypernasality after surgical maxillary advancement is not influenced by the cleft type. These results are important for the clinical practice, since they provide information to the multidisciplinary team and their patients concerning the risks of worsening of the velopharyngeal mechanism, and the possible therapies for speech symptoms. Investigations have been conducted at the Laboratory of Physiology of the Hospital for Rehabilitation of Craniofacial Anomalies of the University of São Paulo in an attempt to identify the factors that may lead individuals with CLP to present appearance or worsening of hypernasality after MA.

Future studies may be conducted aiming to verify the application and effectiveness of complementary therapies after primary palate repair in individuals with poor midface growth, such as maxillary expansion combined to orthodontic treatment or bone-anchored maxillary protraction, aiming to minimize the impact of surgical maxillary advancement on speech in cases in need of this procedure.

# CONCLUSION

The present results demonstrated that the cleft type has no influence on the appearance of hypernasality after surgical maxillary advancement, despite the different anatomical conditions of cleft types affecting the palate.

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#### Author contributions

MNLMS main author, responsible for the study, data collection, data analysis and manuscript writing; BMAMA collaboration in data collection; APF collaboration in manuscript revision; IEKT collaboration in manuscript revision; RPY responsible for the project, study design and supervision of stages of accomplishment and manuscript writing.