

Comparative study among cleft patients with velopharyngeal insufficiency treated with speech therapy and pharyngoplasty

Estudo comparativo entre pacientes fissurados portadores de insuficiência velofaríngea tratados com fonoterapia e faringoplastia

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ABSTRACT

Background: Inadequacy of speech and voice due to velopharyngeal insufficiency is a major stigma for cleft patients. Therefore, the study of this clinical condition is essential to improve the development and social relations of children with this ailment. This study aimed to assess alterations in the speech, velopharyngeal sphincter, and larynx of patients with cleft palate and cleft lip and palate who underwent lip closure, palatoplasty, and speech therapy and developed transitory velopharyngeal insufficiency. Furthermore, these patients were compared with cleft palate and cleft lip and palate patients who developed persistent velopharyngeal insufficiency treated with lip closure, palatoplasty, speech therapy, pharyngoplasty, and complementary speech therapy. **Methods:** From June 1997 to May 2002, 132 cleft palate and cleft lip and palate patients aged between 4 years 11 months and 19 years 3 months with transitory velopharyngeal insufficiency and persistent velopharyngeal insufficiency were assessed. After applying inclusion and exclusion criteria, 44 patients, 18 females and 26 males, were divided into 2 groups: group I, 20 patients who underwent lip closure, palatoplasty and speech therapy; and group II, 24 patients who underwent lip closure, palatoplasty, speech therapy, pharyngoplasty, and complementary speech therapy. Speech therapy consisted of articulatory therapy of oral airflow, myofunctional therapy, rapid phonemic acquisition technique, and voice therapy. Surgical treatment consisted of producing a flap from the pharynx posterior wall of the upper pedicle. **Results:** Alterations in speech, the velopharyngeal sphincter, and the larynx were more frequent in group I than in group II. **Conclusions:** Cleft patients with persistent velopharyngeal insufficiency should be treated with pharyngoplasty and complementary speech therapy in order to correct alterations in speech, the velopharyngeal sphincter, and the larynx.

Keywords: Velopharyngeal insufficiency. Larynx. Cleft palate. Humans.

RESUMO

Introdução: A inadequação da fala e da voz decorrente da insuficiência velofaríngea é o principal estigma do paciente fissurado; assim, o estudo dessa condição clínica é fundamental, proporcionando melhor desenvolvimento das crianças e de suas relações sociais. O objetivo do presente estudo é avaliar alterações fonoaudiológicas, do esfíncter velofaríngeo, da laringe de pacientes fissurados palatais e labiopalatais tratados com queiloplastia, palatoplastia e fonoterapia, que desenvolveram insuficiência velofaríngea transitória, e fissurados palatais e labiopalatais, que desenvolveram insuficiência velofaríngea persistente tratados com queiloplastia, palatoplastia, fonoterapia, faringoplastia e fonoterapia complementar. **Método:** No período de junho de 1997 a maio de 2002, foram avaliados 132 fissurados palatais e labiopalatais que desenvolveram insuficiência velofaríngea transitória e insuficiência velofaríngea persistente, com idade entre 4 anos e 11 meses e 19 anos e 3 meses. Observando-se os critérios de inclusão e exclusão, 44 pacientes, sendo 18 do gênero feminino e 26 do gênero masculino, foram divididos em 2 grupos: grupo I, 20 pacientes submetidos a queiloplastia, palatoplastia e fonoterapia; e grupo II, 24 pacientes submetidos a queiloplastia, palatoplastia, fonoterapia, faringoplastia e fonoterapia complementar. O tratamento fonoaudiológico consistiu de terapia articulatória de fluxo aéreo bucal, terapia miofuncional, técnica de aquisição fonêmica rápida e terapia de voz. O tratamento cirúrgico consistiu de confecção de retalho da parede posterior da faringe, de pedículo superior. **Resultados:** As alterações fonoaudiológicas, do esfíncter velofaríngeo e da laringe foram mais frequentes nos pacientes do grupo I, quando comparados aos do grupo II. **Conclusões:** Pacientes fissurados que evoluíram para insuficiência velofaríngea persistente devem ser tratados com faringoplastia e fonoterapia complementar para correção das alterações fonoaudiológicas, do esfíncter velofaríngeo e da laringe.

Descritores: Insuficiência velofaríngea. Laringe. Fissura palatina. Humanos.

Study conducted at the Plastic Surgery Service of Hospital Municipal Infantil Menino Jesus, Department of Otorhinolaryngology and Head and Neck Surgery of Universidade Federal de São Paulo – School of Medicine of São Paulo (UNIFESP/EPM), São Paulo, SP, Brazil.

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INTRODUCTION

The inadequacy of speech and voice arising from velopharyngeal insufficiency is a major stigma for cleft patients. Therefore, the study of this clinical condition is essential to treat the disease, thereby improving the development of children with this ailment and improving their relations with family, friends, in school and in the community.

In order to carry out treatment, it is important to set up a multispecialty team consisting of a plastic surgeon, speech therapist, dental surgeon, and otorhinolaryngologist. Furthermore, pediatric, nutritional, and psychological follow-up are required. The entire team should be oriented with the treatment protocols.

The anatomical structures of the velopharyngeal sphincter will remain unchanged in approximately 20% of patients treated for cleft palate and cleft lip and palate, resulting in total or partial impairment of function, regardless of the correct performance of lip closure, palatoplasty, and speech therapy¹.

Nasofibroscopy is an important exam for diagnosing velopharyngeal insufficiency, making it possible for the patient to use speech therapy for correction of voice and speech changes by direct visualization of the movements of the velopharyngeal sphincter^{2,3}.

The first assessment of the velopharynx by means of rhinoscopy was performed in 1869⁴. It was verified that complete closure did not always occur during the pronunciation of /a/, /o/, and /u/. Furthermore, the formation of a crease in the posterior wall of the pharynx was observed in individuals with cleft palate and cleft lip and palate. This structure was subsequently called the Passavant crease.

In 1973, the velopharyngeal space received was recognized as a real sphincter⁵.

Laryngeal alterations in patients with cleft palate and cleft lip and palate were studied in 1969, revealing implications for future studies. Children with a moderately suitable velopharyngeal mechanism may produce compensatory movements during speech to counterbalance velopharyngeal insufficiency, causing laryngeal alterations⁶.

In 1961, the velopharyngeal sphincter was studied in normal patients by means of cineradiographies, and synchronized speech was studied by means of spectrography. The author of this study considered 20 mm² as the maximum area for velopharyngeal competence⁷.

In 1973, a pharyngoplasty technique that used a myomucosal flap of the upper pedicle of the pharynx upper wall was described⁸. The control of spaces lateral to the flap was possible with the aid of a vesical probe N° 14 introduced in each nostril, generating a hole in each side of the flap and the lateral wall of the pharynx with an area of 10 mm², totaling 20 mm². Therefore, wide flaps were obtained in patients whose velopharyngeal space diameter was large whereas narrow flaps were obtained in patients with small diameters.

Flaps from the nasal plane of the uvula and soft palate were associated for coverage of the pharyngeal flap open area.

In 2005, a technique that associated the intravelar veloplasty^{9,10} to the inclusion of the upper part of the posterior pillars of amygdalin cavities and of the palatopharyngeal muscles¹¹ was published. New techniques continue to be developed for velopharyngeal insufficiency correction¹².

This paper aimed to assess phono-articulatory alterations of the velopharyngeal sphincter and larynx in cleft patients with velopharyngeal insufficiency treated with speech therapy and who underwent lip closure and palatoplasty. Furthermore, the results from these patients were compared with results from patients who developed persistent velopharyngeal insufficiency treated with pharyngoplasty and complementary speech therapy.

METHODS

In the period from June 1997 to May 2002, 132 patients with transforamen cleft palate and cleft lip and palate were assessed. The patients in this group ranged in age from 4 years 11 months to 19 years 3 months and consisted of 54 females and 78 males. The patients had velopharyngeal insufficiency and were from the outpatient service of the Plastic Surgery of Hospital Municipal Infantil Menino Jesus (São Paulo, SP, Brazil).

Inclusion Criteria

The study included patients who underwent surgical treatment for cleft closure according to the following techniques:

- in cases of unilateral transforamen cleft lip and palate, lip closure was performed by advancement and rotation of flaps in a single procedure¹³;
- in cases of bilateral transforamen cleft lip and palate, lip closure was performed in three procedures¹⁴;
- in cases of unilateral and bilateral transforamen cleft lip and palate, palatoplasty¹⁵ was associated to the vomerine flap¹⁶ in the hard palate and to intravelar veloplasty^{9,10} in the soft palate;
- in cases of isolated cleft palate, palatoplasty¹⁵ was always associated with intravelar veloplasty^{9,10}.

Exclusion Criteria

The following patients and conditions were excluded from the study:

- patients who underwent secondary palatoplasty and/or the presence of a fistula was detected;
- patients with palatal or lip and palate fistula associated with a genetic syndrome;
- other malformations and facial trauma;
- associated diseases;
- low intelligence quotient according to age;
- loss of or altered hearing function.

After observing the inclusion and exclusion criteria, 44 patients were selected of which 18 were female and 26 were male (Table 1).

Patient assessment was performed according to the sequence described below.

1) Speech assessment:

- assessment was performed through spontaneous and stimulated speech;
- voice assessment was performed through hearing-perceptual analysis;
- assessment of the soft palate at rest and at motion was performed during the pronunciation of the vowel /a/.

In voice and speech assessments, which were performed by two speech therapists, the presence or absence of hypernasality, nasal air escape, and articulatory disorders was verified.

Instrumental Assessment of the Velopharyngeal Sphincter and Larynx

Velopharyngeal sphincter assessment was performed by observing the following protocol¹⁷: the patients slowly pronounced the vowels /a/, /e/, /i/, /o/, and /u/ and the consonant /s/ and slowly counted from 1 to 10. Furthermore, the patients pronounced the following sentences: “papai fez a pipa” (daddy made the kite), “Kiki gosta de chá” (Kiki likes tea), “Juju saiu cedo” (Juju left early), and “o cachorro chegou da chuva” (the dog arrived from the rain).

The patients were asked to produce the sound of long /i/ and /é/ while the larynx was examined.

All examinations were recorded on VHS tapes.

The examinations were carried out at the Institute of Otorhinolaryngology and at the Discipline of Pediatric Otorhinolaryngology of UNIFESP/EPM (São Paulo, SP, Brazil).

The following aspects were assessed in the velopharyngeal sphincter examination: closure in deglutition, extension

of lateral wall movements, the presence of a crease in the posterior wall of the pharynx and its level against the velar closure, and adenoid hypertrophy in relation to the velopharyngeal space.

In the larynx examination, we sought to observe the presence of anteroposterior or median constriction, triangular and fusiform glottic apertures, and the presence or absence of vocal nodules.

For comparison purposes, the studied population was divided in two groups:

- group I, consisting of 20 patients, included 8 with cleft palate who underwent palatoplasty and speech therapy and 12 with transforamen cleft lip and palate who underwent lip closure, palatoplasty, and speech therapy – the speech treatment for the correction of hypernasality and articulatory disorders involved articulatory therapy of buccal air flow¹⁸, myofunctional therapy¹⁹, the fast phonemic acquisition technique¹⁸, and voice therapy; and
- group II, consisting of 24 patients, included 7 with cleft palate who underwent palatoplasty and speech therapy and 17 with transforamen cleft lip and palate who underwent lip closure, palatoplasty, and speech therapy and developed persistent velopharyngeal insufficiency treated with pharyngoplasty and complementary speech therapy.

The surgical treatment performed for correcting hypernasality involved the production of a flap of the pharynx posterior wall of the upper pedicle while observing the control of spaces between the flap and the lateral walls of the pharynx^{7,20,21} (Figures 1 to 10).

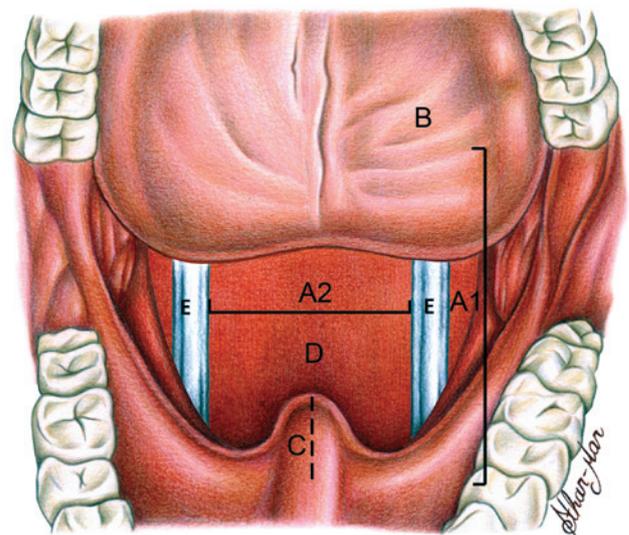


Figure 1 – Marking the width and length of the pharyngeal flap. A1 = flap length; A2 = flap width; B = tongue; C = uvula; D = posterior wall of the pharynx; E = catheter.

Table 1 – Transforamen cleft palate and cleft lip and palate patients with velopharyngeal insufficiency listed according to the type of cleft and gender.

Cleft type	Gender			
	Female		Male	
	n	%	n	%
FP	10	55.6	5	19.2
FLPT/D	1	5.5	4	15.4
FLPT/E	1	5.5	9	34.6
FLPT/B	6	33.4	8	30.8
Total	18	100	26	100

FLPT/B = bilateral transforamen cleft lip and palate; FLPT/D = right transforamen cleft lip and palate; FLPT/E = left transforamen cleft lip and palate; FP = cleft palate; n = number of patients.

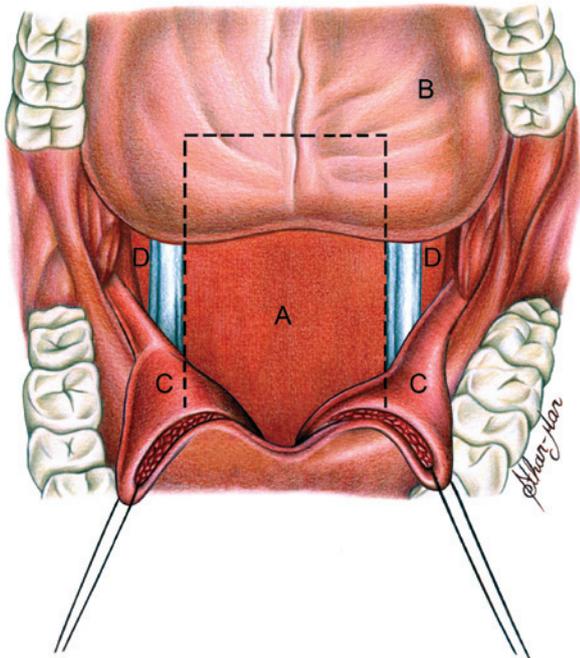


Figure 2 – Layout of the pharyngeal flap. A = flap; B = tongue; C = uvula; D = posterior wall of the pharynx.

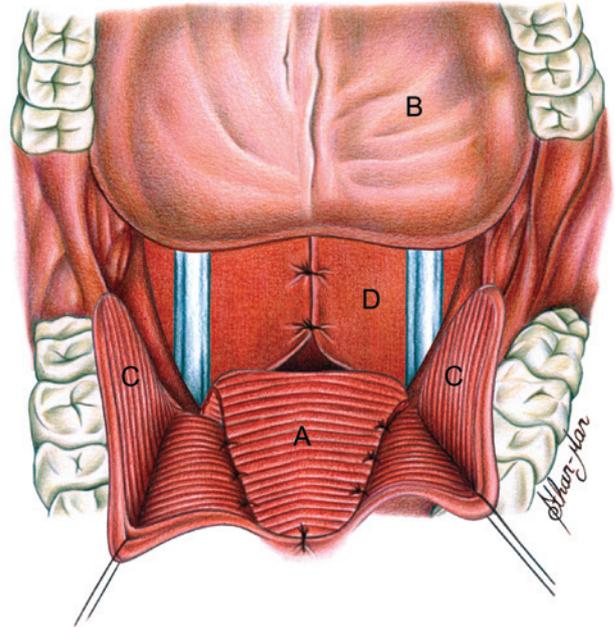


Figure 4 – Suture of the pharyngeal flap in the soft palate. Production of flaps of the nasal plane of the uvula. A = flap; B = tongue; C = uvula; D = posterior wall of the pharynx.

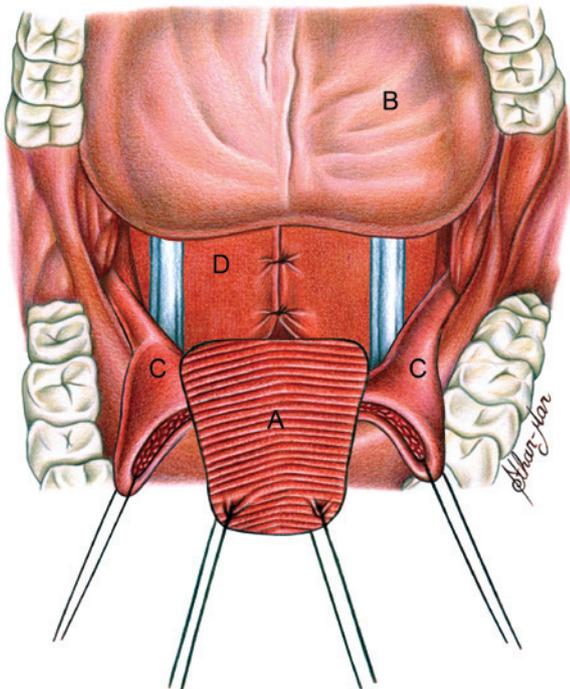


Figure 3 – Incision of the uvula and soft palate and pharyngeal flap raising. A = flap; B = tongue; C = uvula; D = posterior wall of the pharynx.

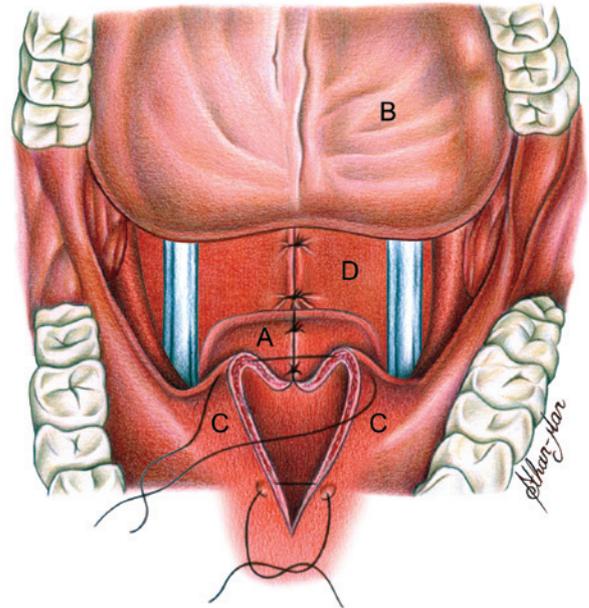


Figure 5 – Suture of uvula flaps over the pharyngeal flap. A = flap; B = tongue; C = uvula; D = posterior wall of the pharynx.

Groups I and II were compared by means of speech and pharyngolaryngoscopic assessment with the purpose of

observing possible improvements in hypernasality, nasal air escape, articulatory disorders, alterations of the velopharyngeal sphincter, and glottic alterations that were observed before the treatment.

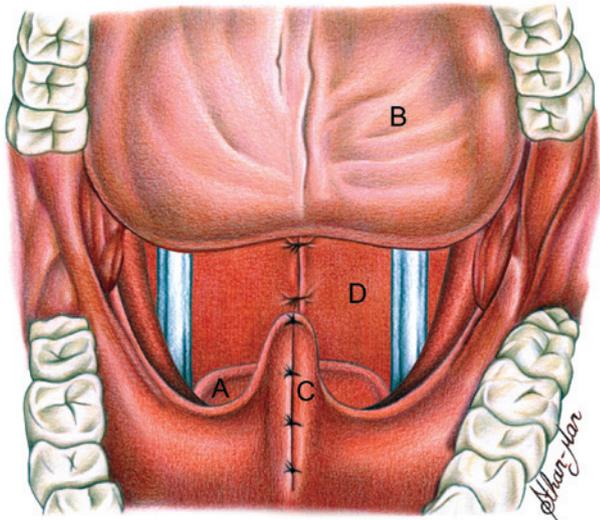


Figure 6 – Suture of the oral plane of the uvula. A = flap; B = tongue; C = uvula; D = posterior wall of the pharynx.

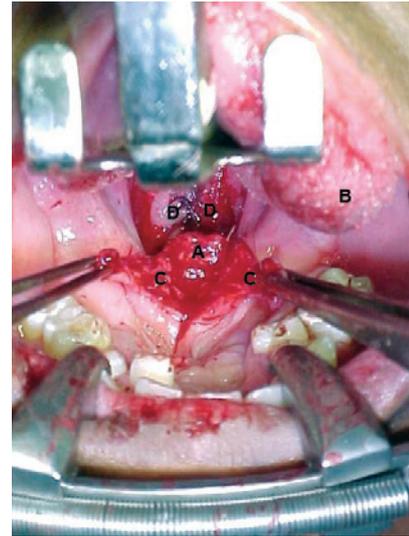


Figure 8 – Surgical procedure. Suture of the pharyngeal flap in the soft palate. A = pharyngeal flap sutured in the soft palate; B = tongue; C = flaps of the nasal plane of the uvula; D = posterior wall of the reconstructed pharynx.



Figure 7 – Surgical procedure. Marking the flap of the pharynx posterior wall. A = marked pharyngeal flap, where length corresponds to the distance marked from the soft palate to the posterior wall of the pharynx; B = tongue; C = marked uvula and soft palate.

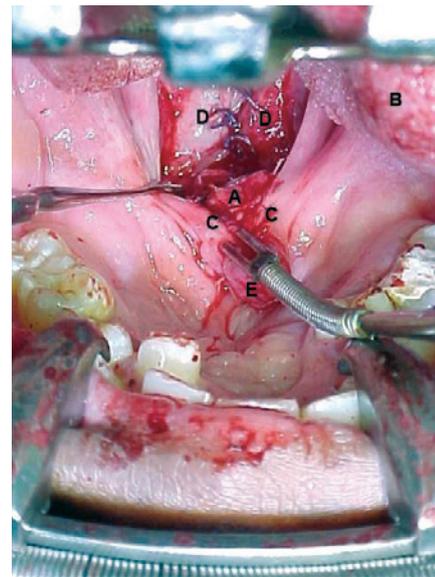


Figure 9 – Surgical procedure. Coverage of the open area of the pharyngeal flap. A = pharyngeal flap sutured in the soft palate; B = tongue; C = flaps of the nasal plane of the uvula, sutured over the open area of the pharyngeal flap; D = posterior wall of the reconstructed pharynx; E = oral plane of the uvula.

The research plan was submitted and approved by the Ethics Committee of UNIFESP/EPM, and an informed consent form was signed by a parent or guardian.

For analysis, we used the chi-square test or the exact test of Fisher²² for 2×2 tables, with the purpose of comparing both groups with regard to the presence of each of the mentioned alterations.

In all tests, the significance level was established at 0.05 or 5%, and the significant values are marked with an asterisk.

RESULTS

The chi-square test²² demonstrated significantly larger frequencies of alterations of speech, the velopharyngeal sphincter, and the larynx in group I (Tables 2 to 4).

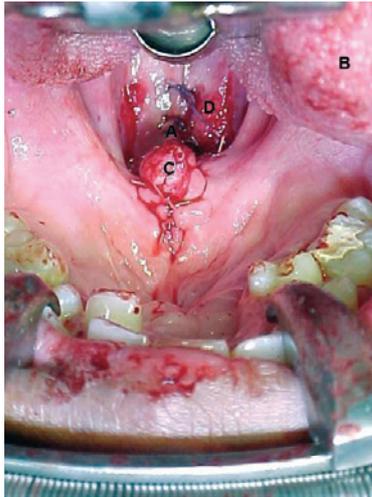


Figure 10 – Surgical procedure. Reconstruction of the uvula oral plane. A = pharyngeal flap covered by flaps of the uvula nasal plane; B = tongue; C = reconstructed uvula; D = posterior wall of the reconstructed pharynx.

Table 2 – Transforamen cleft palate and cleft lip and palate patients with velopharyngeal insufficiency in groups I and II listed according to the presence of speech alterations.

Speech alterations			
Groups	Yes	No	Total
I	11 (55%)	9 (45%)	20 (100%)
II	6 (25%)	18 (75%)	24 (100%)
Total	17	27	44
X ² calculated = 4.14 (P < 0.05)		X ² critical = 3.84 (P < 0.05)	

DISCUSSION

It is difficult to identify the incidence of cleft palate and cleft lip and palate in Brazil because there is no adequate census. It is important to highlight that the incidence tends to increase either by intensification of environmental factors that determine the deformity or by addition of the following factors: (a) decrease of perinatal mortality due to improved puericulture and pediatric techniques that ensure better care for the cleft patient; (b) mortality decrease during the surgical procedures due to better equipment, drugs, and anesthetic techniques; (c) better pre-natal care; and (d) better quality of the surgical technique. These factors result in the integration of cleft patients into society, increasing the possibility that they will marry among themselves and genetically transmit the deformity.

The treatment of cleft lip and palate aims to obtain a result that enables the normal anatomical and functional

Table 3 – Transforamen cleft palate and cleft lip and palate patients with velopharyngeal insufficiency in groups I and II listed according to the presence of velopharyngeal sphincter alterations.

Velopharyngeal sphincter alterations			
Groups	Yes	No	Total
I	10 (50%)	10 (50%)	20 (100%)
II	4 (16.7%)	20 (83.3%)	24 (100%)
Total	14	30	44
X ² calculated = 5.59 (P < 0.02)		X ² critical = 3.84 (P < 0.02)	

Table 4 – Transforamen cleft palate and cleft lip and palate patients with velopharyngeal insufficiency in groups I and II listed according to the presence of larynx alterations.

Larynx alterations			
Groups	Yes	No	Total
I	11 (55%)	9 (45%)	20 (100%)
II	6 (25%)	18 (75%)	24 (100%)
Total	17	27	44
X ² calculated = 4.14 (P < 0.02)		X ² critical = 3.84 (P < 0.02)	

development of each patient. The inadequacy of speech and voice derived from velopharyngeal insufficiency is a major stigma for cleft patients. Therefore, the study of this clinical condition is essential in order to treat this disease, thereby improving the development of children with this condition and improving their relationships with family, friends, in school, and the community.

Following the description of the anatomical and functional bases of velopharyngeal occlusion by Passavant⁴ in 1869, and the suggestion that speech can be improved by fixation of the soft palate in the posterior wall of the pharynx, several techniques have been described for this process. In 1973, Hogan⁸ described a technique that used a myomucosal flap of the upper pedicle of the pharynx posterior wall. The modification introduced by Hogan was important because it achieved control of the spaces between the flap and lateral walls of the pharynx by introducing a vesical probe N° 14 in each nostril. Therefore, each orifice lateral to the flap generated an area corresponding to 10 mm², totaling 20 mm². In 1961, Bjork⁷ studied the velopharyngeal sphincter of normal patients by means of cineradiographies and synchronized speech and reported that 20 mm² is the maximum area for velopharyngeal competence. Another modification introduced by Hogan was coverage of the open area of the pharyngeal flap using flaps from the nasal plane

of the uvula and soft palate. These modifications allowed obtainment of wide flaps for correction of the velopharyngeal insufficiency in patients with a velopharyngeal space with a large diameter and obtainment of narrow flaps in patients with a small diameter.

The results achieved in several world centers that specialize in the treatment of patients with cleft palate and cleft lip and palate are superior in patients who undergo palatoplasty before 12 years of age. At the Hospital Municipal Infantil Menino Jesus, palatoplasties are frequently carried out after 18 months of age and, in some cases, after 24 months of age. This range is mainly because of the time that elapses between childbirth and the first consultation.

In group I, the results of patients who underwent palatoplasty at up to 2 years of age were modestly better (37.5% success rate) than in patients who underwent palatoplasty after 2 years of age (33.3% success rate). In group II, the results were much better under the same age conditions (80% versus 57.1%). These outcomes could not be analyzed statistically because of the dimension of the sample. We observed an 81.8% success rate in patients that underwent pharyngoplasty between 5 and 10 years of age and a 53.8% success rate in patients that underwent this procedure at more than 10 years of age. Statistical analysis could not be performed because of the dimension of the sample.

At the Hospital Municipal Infantil Menino Jesus, the treatment of cleft patients is carried out by a multispecialty team; this is consistent with the approach of Centers of Cleft Treatment all over the world.

The adaptation of movements of the pharynx lateral wall to the pharyngeal flap was described in 1999. A statistically significant adaptation of adduction of the pharynx lateral walls was observed after the pharyngoplasty²³.

In group II, 20 (83.33%) patients had coronal-type closure and 4 (16.67%), had circular type closure. Following treatment, coronal-type closure remained in 14 (58.34%) patients, circular-type closure remained in 5 (20.83%) patients, and circular-type closure with a Passavant ridge was observed in 5 (20.83%) patients. These results are consistent with a study conducted in 1980²⁴. Patients who undergo pharyngoplasty are able to change the movement of the lateral walls, and in some cases, the configuration of these movements may be altered.

For the purpose of statistical analysis, the cases that evolved without hypernasality, nasal air escape, articulatory disorders, vocal crease nodules, constriction, and glottic aperture and with suitable closure of the velopharyngeal sphincter were considered successful, whereas the presence of one of these undesired alterations was considered a failure.

The presence of these alterations was compared in both groups and the chi-square test²² revealed that the frequencies of all alterations were significantly smaller in group II.

The incidence of vocal disorders in cleft lip and palate and cleft palate pre-school age patients has been studied. Children

with moderate or severe hypernasality were statistically proven to have medium hypernasality or normal resonance, especially those children that produced the coup de glotte more often²⁵.

Laryngeal alterations occurred in both group I and group II patients, indicating the need for larynx examination in all patients with velopharyngeal insufficiency.

Prospective studies will be necessary to determine if there is causal relationship between abnormalities of the velopharyngeal sphincter and laryngeal alterations in individuals.

CONCLUSIONS

Cleft patients who undergo lip closure, palatoplasty, and speech therapy and develop persistent velopharyngeal insufficiency should be treated with pharyngoplasty and complementary speech therapy in order to correct alterations in speech, the velopharyngeal sphincter, and the larynx.

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