

Surgical Treatment of Submucous Cleft Palate: A Comparative Trial of Two Modalities for Palatal Closure

**Antonio Ysunza, M.D., Sc.D., Ma. Carmen Pamplona, B.S., Mario Mendoza, M.D.,
Fernando Molina, M.D., Patricia Martínez, M.D., Manuel García-Velasco, M.D., and Nicolas Prada, M.D.**

Mexico City, Mexico

Submucous cleft palate is a congenital malformation with specific clinical and anatomical features. It can be present with or without velopharyngeal insufficiency. Surgical treatment of this malformation is indicated only when velopharyngeal insufficiency has been demonstrated. This article compares two modalities of surgical treatment for submucous cleft palate. The first includes a minimal incision palatopharyngoplasty, as described in a previous report. The second combines the first technique with additional individualized velopharyngeal surgery (individualized pharyngeal flap or sphincter pharyngoplasty) performed simultaneously. The individualized part of the procedure was selected and performed according to the findings of videonasopharyngoscopy and multiview videofluoroscopy, as reported previously. Two hundred and three patients with submucous cleft palate were studied from 1990 to 1999. Videonasopharyngoscopy and multiview videofluoroscopy demonstrated velopharyngeal insufficiency in 72 patients, who were randomly divided into two groups. Those in group 1 ($n = 37$) underwent a minimal incision palatopharyngoplasty. Patients in group 2 ($n = 35$) also underwent that procedure but simultaneously received individualized pharyngeal flap or sphincter pharyngoplasty, according to the findings of videonasopharyngoscopy and multiview videofluoroscopy. The median age of the patients from both groups was not significantly different ($p > 0.5$). The frequency of residual velopharyngeal insufficiency after palatal closure was not significantly different in both groups of patients (14 percent versus 11 percent; $p > 0.5$). The mean size of the gap at the velopharyngeal sphincter during speech was not significantly different in both groups of patients before surgery (23 percent versus 22 percent; $p > 0.5$). After the surgical procedures, there was a nonsignificant difference between both groups of patients in mean residual size of the gap in cases of velopharyngeal insufficiency (7 percent versus 8 percent; $p > 0.5$). It seems that minimal incision palatopharyngoplasty is a safe and reliable procedure for palatal closure in patients with submucous cleft palate. The use of additional individualized velopharyngeal surgery performed simultaneously did not seem to decrease the frequency of residual velopharyngeal insufficiency.

Moreover, the residual size of the gap at the velopharyngeal sphincter was not significantly reduced when an additional surgical procedure was performed simultaneously with palatal closure. (*Plast. Reconstr. Surg.* 107: 9, 2001.)

Velopharyngeal insufficiency refers to abnormalities of the velopharyngeal sphincter involving the velum and/or pharyngeal walls.¹ Although the disorder is commonly seen in patients with cleft palate and therefore is diagnosed in early evaluations, submucous cleft palate is overlooked in some individuals with velopharyngeal insufficiency. In this latter population, the diagnosis might be delayed until speech development allows detection by skilled personnel.^{2,3} There is a distinction between the longitudinal observation of the complete cleft palate patient and the usual determination of submucous cleft palate with velopharyngeal insufficiency when a child starts school. Submucous cleft palate was defined by Calnan as a triad of physical findings, including a notched posterior hard palate and zona pellucida (midline muscle separation) and a bifid uvula.⁴ The incidence of submucous cleft palate in the general population is not known; reports range from 1:10,000 to 1:200.^{2,5-7} Submucous clefts of the palate continue to present a diagnostic and therapeutic challenge. Clinicians may see a disproportionately higher percentage of individuals with this disorder among patients with speech difficulties and chronic ear infections. The actual frequency of abnormal speech among individuals

with isolated submucous cleft palate (i.e., non-syndromic) is actually quite low,^{6,7} probably under 5 percent. Once diagnosed, the question is whether speech therapy alone will resolve the velopharyngeal insufficiency or if surgery is required. It is important to obtain an accurate assessment of speech before a therapeutic decision is made. This assessment requires the cooperation of the patient and usually cannot be adequately completed until the patient is approximately 3 or 4 years of age.^{1,2} Diagnosis of submucous cleft palate can be made on the basis of the classic triad described by Calnan. However, some patients with the disorder do not have the complete triad. Thus, diagnosis is frequently made by videonasopharyngoscopy.

Videonasopharyngoscopy provides visualization of the velopharyngeal sphincter during speech. This procedure can define the absence of musculus uvulae bulge when a patient is evaluated for "noncleft" velopharyngeal insufficiency. Besides these physical findings, the patient usually is referred for evaluation of the velopharyngeal insufficiency or other functional problems, such as compensatory articulation disorder. This disorder, which is considered to be one of the signs and symptoms of velopharyngeal insufficiency, occurs during the attempt to articulate fricative and explosive sounds by stopping, starting, or otherwise manipulating the air column during speech lower in the vocal tract, such as at the glottis or hypopharynx.⁸ Perceptual assessment can confirm hypernasality, but the standard evaluation for velopharyngeal insufficiency includes flexible videonasopharyngoscopy and multiview videofluoroscopy. These procedures provide the best direct assessments to help plan and direct the optimal treatment of velopharyngeal insufficiency.⁹

Normality of the final speech results in all patients with velopharyngeal insufficiency depends on articulation as much as normal resonance balance. Nasal resonance is corrected by physical management of the velopharyngeal sphincter (surgery or prosthetic appliances). Compensatory articulation disorder associated with velopharyngeal insufficiency requires speech therapy because of the dysfunction not only of the velopharyngeal sphincter but also of the entire vocal tract.⁸ The goal in treating velopharyngeal insufficiency is to restore a functional seal between the nasopharynx and oropharynx so that normal articulation of speech occurs. Several options are available.

Good results have been reported with different techniques, including the Wardill push-back procedure.¹⁰⁻¹² In addition, individualized velopharyngeal surgery is commonly performed when palatal closure fails to completely correct velopharyngeal insufficiency.¹³ Individualized surgery includes customized pharyngeal flaps and sphincter pharyngoplasties performed according to the findings of videonasopharyngoscopy and multiview videofluoroscopy, as reported previously.^{8,14,15}

This article compares two modalities of the surgical correction of submucous cleft palate. The first is a minimal incision palatopharyngoplasty, as described earlier.¹² The second includes the first technique plus an additional surgical procedure performed simultaneously; that is, either a pharyngeal flap or a sphincter pharyngoplasty, depending on the findings of videonasopharyngoscopy and multiview videofluoroscopy, as reported previously.^{8,14,15}

MATERIALS AND METHODS

Sample size was calculated at an alpha 95 percent confidence interval, and a beta power of 80 percent for a comparative study of the two groups. The frequency of velopharyngeal insufficiency in cases of submucous cleft palate, and the frequency of residual velopharyngeal insufficiency after palatal closure during 2 previous years at the cleft palate clinic, were considered. These data indicated at least 32 patients for inclusion in each group.¹⁶

A prospective study was completed of patients diagnosed with submucous cleft palate at the cleft palate clinic of the Hospital Gea Gonzalez at Mexico City. All patients diagnosed with submucous cleft palate from January of 1990 to July of 1999 were studied. During this period, 203 patients who presented with submucous cleft palate without prior treatment were evaluated at the cleft palate clinic. The protocol was approved by the Research Committee and the Bioethics Committee of the Hospital Gea Gonzalez. All parents and legal guardians were counseled before the patients were included in the study group. The methodology of the study and the surgical procedures were carefully explained to all parents and legal guardians. The parents and legal guardians of all selected patients agreed to participate in the study.

To qualify for the subject group for this study, the patients had to meet the following criteria:

1. A diagnosis of nonsyndromic submucous cleft palate, with no other medical condition. To be included in the study, all patients had to have at least two of the three clinical triads as determined by oral examination and/or videonasopharyngoscopy and multiview videofluoroscopy. All three of Calnan's criteria were not required to be present for the diagnosis of submucous cleft palate, because as reported previously,² inconsistencies in their presence should be considered as a variant of submucous cleft palate, much like the variants in the Pierre-Robin sequence. Although a bifid uvula may be the first indication that the patient may have submucous cleft palate, it alone is not a reliable sign of the diagnosis.

2. Velopharyngeal insufficiency with or without compensatory articulation disorder as demonstrated by phoniatric assessment, videopharyngoscopy, and multiview videofluoroscopy.

3. Chronological age between 4 and 8 years of age at the time of selection for the study.

4. Normal hearing as demonstrated by conventional pure-tone audiometry.

5. Language development within reference limits as demonstrated by a battery of age-appropriate standardized language tests.¹⁷

A total of 72 patients met the criteria for inclusion in the study group. The remaining 131 patients with submucous cleft palate included those without velopharyngeal insufficiency and those with associated anomalies, delayed linguistic development, and hearing loss.

Twenty-nine patients showed velopharyngeal insufficiency and associated compensatory articulation disorder as demonstrated by phoniatric assessment, videonasopharyngoscopy, and multiview videofluoroscopy. These patients received speech therapy to correct compensatory articulation disorder until placement of articulation was normal during connected speech even when hypernasality was present. At the end of the speech therapy period, all patients were independently examined by two speech pathologists. The patients did not continue the study protocol until both pathologists were convinced that the compensatory articulation disorder had been corrected. Total time of speech therapy was considered to be the time from onset therapy until the normalization of placement of articulation during connected speech even when hypernasality was present.

After the speech therapy period, the 29 pa-

tients underwent additional videonasopharyngoscopy and multiview videofluoroscopy to record data of the velopharyngeal sphincter without the influence of compensatory articulation disorder. Finally, these 29 cases were included within the whole study group of 72 cases.

The 72 patients were randomly divided into two groups: 37 in group 1 (control) and 35 in group 2 (active). Patients in group 1 were operated on by using a minimal incision palatopharyngoplasty, as described earlier.¹² Those in group 2 received the same procedure plus simultaneous individualized velopharyngeal surgery according to the findings of videonasopharyngoscopy and multiview videofluoroscopy. The size and form of the gap, lateral pharyngeal wall motion, and level of maximum displacement of the velopharyngeal sphincter were considered as criteria for individualizing the surgical procedure.^{8,13} All surgeries were performed by the same team, which included two of the present authors (M. M. and F. M.).

Three months after the surgical procedures, all patients underwent additional phoniatric evaluation. A blind procedure was used whereby all analyses were independently conducted by two speech pathologists with 10 years of experience evaluating and treating cleft palate patients. Only perceptual evaluations were used to keep the examiners blind to the surgical procedures that had been performed. Post-operative videonasopharyngoscopy and multiview videofluoroscopy were also performed 3 months after the surgical procedures. All of these evaluations were independently analyzed by two speech pathologists who were trained in the procedures. The presence or absence of velopharyngeal insufficiency and the size of the defect at the velopharyngeal sphincter during speech were determined. Concordance between each pair of examiners was evaluated. When differences occurred, each case was discussed by the examiners until an agreement was reached.

The results from both groups of patients were compared. Age was considered as one dimensional variable and was analyzed by a Mann-Whitney rank sum test. Another dimensional variable was the size of the gap as observed by videonasopharyngoscopy and multiview videofluoroscopy, which was analyzed by Student's *t* test. Velopharyngeal insufficiency was considered as a binary variable (yes or no) and was analyzed with a Fisher's exact test. For

each variable, an alpha value of 0.05 was selected for considering the results as stochastically significant.¹⁶

RESULTS

All patients had at least two of the three physical findings of the triad; 70 percent of the patients had all three criteria. Ninety-eight percent of the patients had a palpable notch in the posterior hard palate. Eighty percent had an identifiable midline muscle diastasis, and 85 percent presented with a bifid uvula. Videonasopharyngoscopy demonstrated absence of musculus uvulae in 94 percent of the patients.

The age of the patients in group 1 (control, $n = 37$) ranged from 4 years to 7 years, 4 months (median: 5 years, 5 months). In group 2 (active, $n = 35$) ages ranged from 4 years to 7 years, 7 months (median: 5 years, 3 months). A Mann-Whitney rank sum test revealed a non-significant difference in age between both groups ($p > 0.5$).

The total time of speech therapy in the 29 patients who presented with velopharyngeal insufficiency and compensatory articulation disorder ranged from 38 to 95 months (mean \pm SD, 62.2 ± 22.5). Although 90 percent of the patients demonstrated an improvement in velopharyngeal movements and a reduction in gap size after correction of compensatory articulation, in none of the patients was velopharyngeal insufficiency corrected by speech therapy. The opinions of the two examiners for the preoperative videonasopharyngoscopy and multiview videofluoroscopy coincided in 98 percent of the cases.

Closure patterns of the velopharyngeal sphincter during speech were evenly distributed between the two treatment groups. Forty percent of the patients showed a coronal pattern, 50 percent showed a circular pattern, and 10 percent showed Passavant's ridge. The gap size of the velopharyngeal closure during speech as observed preoperatively by videonasopharyngoscopy and multiview videofluoroscopy was not significantly different for both groups (23.4 ± 11.3 percent in group 1; 22.9 ± 10.4 percent in group 2; $p > 0.5$).

Of the patients who had additional individualized velopharyngeal surgery (group 2, active), 3 received sphincter pharyngoplasty and 32 underwent pharyngeal flap surgery. The surgeons, phoniatrist, and speech pathologists participated in all cases, as described previously.^{8,13}

Opinions of the two examiners for the postoperative phoniatric assessment coincided in 95 percent of the cases. When postoperative results of the videonasopharyngoscopy and multiview videofluoroscopy were examined, the two examiners agreed in 98 percent of the cases.

The success rate for correcting velopharyngeal insufficiency after the surgical procedures was not significantly different for both groups ($p > 0.05$). Five patients (14 percent) from group 1 and four patients (11 percent) from group 2 demonstrated postoperative velopharyngeal insufficiency. Complete closure of the velopharyngeal sphincter was achieved in 32 patients (86 percent) from group 1 and in 31 patients (89 percent) from group 2. After surgery, when the nine patients with residual velopharyngeal insufficiency were considered, the gap size of velopharyngeal closure during speech as observed by videonasopharyngoscopy and multiview videofluoroscopy was not significantly different for both groups (7.4 ± 3.2 percent for group 1; 8 ± 4.1 percent for group 2; $p > 0.5$).

DISCUSSION

The results of this study suggest that the prevalence of velopharyngeal insufficiency was not reduced when additional, individualized velopharyngeal surgery was simultaneously performed with palatal closure by using minimal incision palatopharyngoplasty. Moreover, minimal incision palatopharyngoplasty seems to be a useful tool in treating velopharyngeal insufficiency. It should be pointed out that although the surgery was individualized in all cases in group 2, in 3 patients with persistent velopharyngeal insufficiency after surgery (in whom a wide flap had been prescribed), videonasopharyngoscopy and multiview videofluoroscopy showed moderately narrow flaps. The other patient with persistent velopharyngeal insufficiency underwent sphincter pharyngoplasty. In other words, the surgery was unsuccessful in terms of meeting the prescribed surgical goal in these four cases. Nonetheless, the size of the gap at the velopharyngeal sphincter and the hypernasality were reduced in all cases.

It is generally accepted that surgical treatment of submucous cleft palate is indicated only in the presence of velopharyngeal insufficiency. As demonstrated by several studies, not all patients with a submucous cleft palate develop velopharyngeal insufficiency.^{2,18} There-

fore, surgical intervention should wait until speech and language development are sufficient to allow a thorough evaluation of velopharyngeal function. The incidence of velopharyngeal insufficiency in a large series of patients with submucous cleft palate has been reported to be as low as 5 percent.^{2,5-7} However, other studies report velopharyngeal insufficiency in more than 50 percent of the cases of submucous cleft palate.^{2,18} In this study, 72 of 203 patients with submucous cleft palate demonstrated velopharyngeal insufficiency. The patients in this study, and the individuals who referred them, may well represent a biased group in that the patients were sent to the cleft palate clinic. This factor may have contributed to the high percentage (35 percent) of velopharyngeal insufficiency in this series. Moreover, this finding is also consistent with the knowledge that it is primarily patients with symptomatic velopharyngeal insufficiency who are referred to craniofacial centers or cleft palate clinics for evaluation and treatments.

Although the results from this study seem to support minimal incision palatopharyngoplasty as a safe and reliable procedure for treating patients with complete or submucous clefts of the palate,^{8,19} it is our intention to present it as the procedure of choice for treating submucous cleft palate. It is not that minimal incision palatopharyngoplasty is as successful as an individualized pharyngeal flap; rather, it seems that the pharyngeal flap is as unsuccessful as minimal incision palatopharyngoplasty. Nor is it our intention to present the latter technique as the "new" procedure for palatal closure in patients with submucous clefts. As mentioned herein, other options are available. Several authors report similarly good results with procedures such as Furlow's palatoplasty, intravelar veloplasty, and the Wardill push-back procedure.^{2,18,20,21} Nonetheless, the results from this study suggest that in cases of submucous cleft palate, the use of additional individualized velopharyngeal surgery does not seem to improve the outcome.

It can be said that performing a sphincter pharyngoplasty simultaneously with a minimal incision palatopharyngoplasty presents a considerable risk because of the resulting circular inflammatory process at the velopharyngeal space. However, our experience shows that it is technically possible to perform these procedures simultaneously. Nonetheless, it is abso-

lutely necessary to keep these patients under strict clinical surveillance in the immediate postoperative period. In the 35 cases operated on with the surgical routines mentioned herein, we had no complications such as postoperative bleeding, fistulas, or upper airway obstruction.

None of the parents described having obstructive sleep apnea after surgery. As noted earlier, in the cleft palate clinic of the Hospital Gea Gonzalez, all patients who present with submucous cleft palate routinely undergo videonasopharyngoscopy and multiview videofluoroscopy before palatal closure. When the risk of obstruction is detected, tonsillectomy is performed before velopharyngeal surgery and/or nasopharyngeal tubes are routinely used in the postoperative period.²²

The relatively small number of patients and the homogeneity²³ of the sample included in this study do not allow definite conclusions. However, of those patients having surgery, satisfactory outcome was achieved by a minimal incision palatopharyngoplasty. Moreover, the use of additional individualized pharyngeal flap or sphincter pharyngoplasty does not seem to enhance velopharyngeal function. Thus, it seems appropriate to approach submucous cleft palate with a single procedure for palatal closure. Should this procedure fail to achieve its goal, it would not interfere with the use of other well-known methods for treating secondary velopharyngeal insufficiency, including individualized pharyngeal flap or sphincter pharyngoplasty.

Antonio Ysunza, M.D., Sc.D.
Hospital Gea González
4800 Calzada Tlalpan
México D.F. 14000
amysunza@datasys.com.mx

REFERENCES

1. Shprintzen, R. J., and Golding-Kushner, K. J. Evaluation of velopharyngeal insufficiency. *Otolaryngol. Clin. North Am.* 22: 519, 1989.
2. García-Velasco, M., Ysunza, A., Hernandez, X., and Marquez, C. Diagnosis and treatment of submucous cleft palate. *Cleft Palate J.* 25: 171, 1988.
3. Dixon-Wood, V. L., Williams, W. N., and Seagle, M. B. Team acceptance of specific recommendations for the treatment of VPI as provided by speech pathologists. *Cleft Palate Craniofac. J.* 28: 285, 1991.
4. Calnan, J. Submucous cleft palate. *Br. J. Plast. Surg.* 6: 264, 1954.
5. Kaplan, E. N. The occult submucous cleft palate. *Cleft Palate J.* 12: 356, 1975.

6. Shprintzen, R. I., Schwartz, R. H., Daniller, A., and Hoch, L. Morphologic significance of bifid uvula. *Pediatrics* 75: 553, 1985.
7. Weatherley-White, R. C. A., Sakura, C. Y., Jr., Brenner, L. D., Stewart, J. M., and Ott, J. E. Submucous cleft palate, its incidence, natural history and indications for treatment. *Plast. Reconstr. Surg.* 49: 297, 1972.
8. Pamplona, M., Ysunza, A., Guerrero, M., et al. Surgical correction of velopharyngeal insufficiency with and without compensatory articulation. *Int. J. Pediatr. Otorhinolaryngol.* 34: 53, 1996.
9. Golding-Kushner, K. J., Argamaso, R. V., Cotton, R. T., et al. Standardization for the reporting of nasopharyngoscopy and multiview videofluoroscopy: A report from an International Working Group. *Cleft Palate J.* 27: 337, 1990.
10. Wardill, W. M. The technique of operation for cleft palate. *Br J. Surg.* 25: 117, 1937.
11. Furlow, L. T., Jr. Cleft palate repair by double opposing Z-plasty. *Plast. Reconstr. Surg.* 78: 724, 1986.
12. Mendoza, M., Molina, F., Azzolini, C., and Ysunza Rivera, A. Minimal incision palatopharyngoplasty. *Scand. J. Plast. Reconstr. Surg. Hand Surg.* 28: 199, 1994.
13. Peat, B. G., Albery, E. H., Jones, K., and Pigott, R. W. Tailoring velopharyngeal surgery: The influence of etiology and type of operation. *Plast. Reconstr. Surg.* 93: 948, 1994.
14. Ysunza, A., Pamplona, M. C., Molina, F., Chacón, E., and Collado, M. Velopharyngeal motion after sphincter pharyngoplasty: A videonasopharyngoscopic and electromyographic study. *Plast. Reconstr. Surg.* 104: 905, 1999.
15. Shprintzen, R. J., Lewin, M. L., Croft, C. B., et al. A comprehensive study of pharyngeal flap surgery: Tailor made flaps. *Cleft Palate J.* 16: 46, 1979.
16. Feinstein, A. *Multivariable Analysis*. New Haven: Yale University Press, 1996. Pp. 147-341.
17. Norris, J., and Hoffman, P. *Whole Language Intervention for School Age Children*. San Diego: Singular Publishing Group, Inc., 1996. Pp. 1-311.
18. Seagle, M. B., Patti, C. S., Williams, W. N., and Wood, V. D. Submucous cleft palate: A 10-year series. *Ann. Plast. Surg.* 42: 142, 1999.
19. Ysunza, A., Pamplona, M. C., Mendoza, M., García-Velasco, M., Aguilar, M. P., and Guerrero, M. E. Speech outcome and maxillary growth in patients with unilateral complete cleft lip/palate operated on at 6 versus 12 months of age. *Plast. Reconstr. Surg.* 102: 675, 1998.
20. Gunther, E., Wisser, J. R., Cohen, M. A., and Brown, A. S. Palatoplasty. Furlow's double-reversing Z-plasty versus intravelar veloplasty. *Cleft Palate Craniofac. J.* 35: 546, 1998.
21. Brothers, D. B., Dalston, R. W., Peterson, H. D., and Lawrence, W. T. Comparison of the Furlow double-opposing Z-palatoplasty with the Wardill-Kilner procedure for isolated clefts of the soft palate. *Plast. Reconstr. Surg.* 95: 969, 1995.
22. Ysunza, A., García-Velasco, M., Garcia-Garcia, M., et al. Obstructive sleep apnea secondary to surgery for velopharyngeal insufficiency. *Cleft Palate Craniofac. J.* 30: 387, 1993.
23. Shprintzen, R. J. Fallibility of clinical research. *Cleft Palate Craniofac. J.* 28: 136, 1991.