Frequency of Pharyngoplasty after Primary Repair of Cleft Palate

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Abstract

Background: *The frequency of pharyngoplasty after initial repair of cleft palate is a direct measure of the success or failure of the primary palatal repair with respect to velopharyngeal function. The optimal timing and surgical technique for the repair of cleft palate remain subjects of debate.*

Purpose: *To assess the outcome of various techniques for primary palatal repair, specifically the frequency of secondary pharyngoplasty, and to determine the significance, if any, of certain variables to this outcome.*

Methods: *A pool of 114 patients with cleft lip and palate was compiled from a retrospective analysis of medical records for more than 300 consecutive patients treated over a 15-year period (1980–1995). The review included only patients who had been treated by the same surgeon or by his resident. A 1-stage palatal repair was performed on all patients, in which the hard and the soft palate were closed simultaneously. The following data were collected: patient's sex, patient's date of birth, type of cleft, technique used for initial repair, age at initial repair and date of secondary pharyngoplasty surgery, if performed.*

Results: *The overall frequency of subsequent pharyngoplasty was 25% (28 patients). The rate of secondary surgery was significantly higher for boys (21/63 or 33%) than for girls (7/51 or 14%). There were also significant differences in the rate of secondary pharyngoplasty according to type of cleft: 50% (6/12) for patients with bilateral cleft lip and palate, 44% (7/16) for those with hard and soft cleft palate, 21% (8/38) for those with unilateral cleft lip and palate, 20% (3/15) for those with submucous cleft palate and 12% (4/33) for those with soft cleft palate. Surgical technique for the primary repair (V-Y pushback or von Langenbeck procedure) was not a significant factor in determining the rate of subsequent pharyngoplasty, nor was age at primary repair, although those who underwent primary repair at age 12–14 months were least likely to require pharyngoplasty.*

Conclusion: *In this study the frequency of velopharyngeal insufficiency after 1-stage palatoplasty was consistent with previously reported results. Of interest would be a comparison of 1-stage and 2-stage approaches to primary palate repair in young patients.*

MeSH Key Words: *cleft palate/surgery; cleft palate/complications; velopharyngeal insufficiency*

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Cleft palate occurs when the palatal shelves fail to come fully together and fuse between the eighth and twelfth weeks of embryonic development. The opening that remains permits communication between the nasal passages and the mouth. Clefting varies in severity and often involves the lip. In extreme cases, facial clefts are seen. Cleft lip and palate is one of the most frequent congenital anomalies, and with an overall incidence of roughly 1 in 1,000 North American births,[1] it is the most common craniofacial anomaly. The incidence of cleft palate alone is roughly 1 in 2,000 births.[1]

The ideal surgical approach for management of the cleft palate deformity continues to be a source of controversy. The goal of cleft palate surgery is to repair the defect and allow normal facial growth and speech development. Different treatment approaches have evolved over the years in an attempt to balance facial growth with speech development, esthetic considerations and the child's social needs.

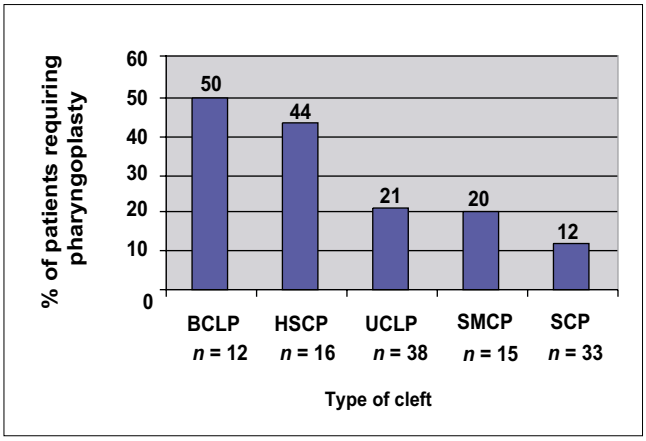

Figure 1: *Need for pharyngoplasty as a function of type of cleft for 114 patients who underwent primary repair of cleft palate by one surgeon between 1980 and 1995. The differences among categories were significant (*p *= 0.030). BCLP = bilateral cleft lip and palate, HSCP = hard and soft cleft palate, UCLP = unilateral cleft lip and palate, SMCP = submucous cleft palate, SCP = soft cleft palate.*

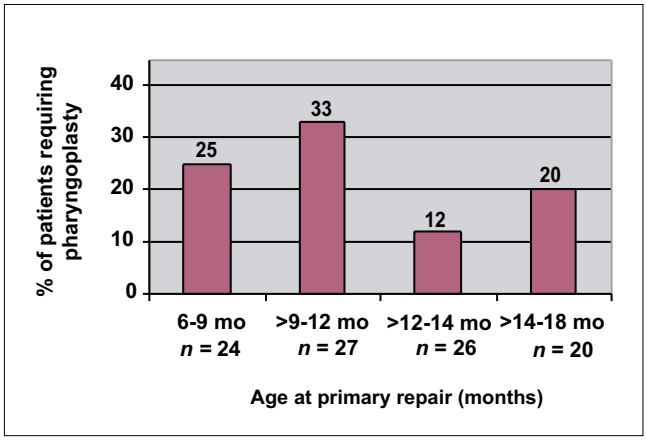

Figure 2: *Need for pharyngoplasty as a function of age at primary repair for 97 patients who underwent primary repair of cleft palate by one surgeon between 1980 and 1995 (patients who underwent primary repair at ages older than 18 months were excluded from this analysis because of other factors that influence the success of operations performed at older ages). The differences were not statistically significant (*p *= 0.29).*

Initial surgical repair of a cleft palate, called palatoplasty, does not always result in normal functioning of the palate. If the soft palate continues to function improperly, the patient may experience speech difficulties because of inability to control the flow of air into the nose. This lack of control of airflow results in hypernasality, whereby the nasal resonance during speech is greater than normal. This problem may be diagnosed as velopharyngeal insufficiency — the inability to completely close the velopharyngeal sphincter during speech. To correct this condition, a secondary surgical procedure known as pharyngoplasty can be performed. Here, the surgeon creates a flap of tissue from the pharynx, which connects the soft palate to the back of the throat. This flap of tissue allows the patient to more easily direct the flow of air through the mouth and away from the nose during speech.

One measure of the success of primary palatal repair is the need for secondary pharyngoplasty. The goal of this study was to determine the frequency of pharyngoplasty after primary palatal repair at the authors' centre. As well, the significance, if any, of certain variables to the patients' outcome was assessed.

Methods

This retrospective study involved a chart review for consecutive patients who underwent primary palatal repair between 1980 and 1995. Only patients who had been treated by the same surgeon or by his resident at the Winnipeg Health Sciences Centre, in Winnipeg, Manitoba, were considered for the study. The patients' charts were accessed through the hospital's medical records department. The analysis was conducted in 2001, and 1995 was selected as the closing year for the eligibility period because it was felt that 6 years from the date of the primary palatal repair to the time of analysis would be sufficient for accurate assessment of speech capabilities and determination of need for secondary pharyngoplasty. From the surgeon's experience, children with velopharyngeal sufficiency at this stage (i.e., 6 years after palatoplasty) are unlikely to experience subsequent insufficiency.

The following data were extracted from the charts: sex, date of birth, type of cleft, technique used for primary repair, age at primary repair and date of secondary pharyngoplasty, if performed. Statistical significance of differences was tested with logistic regression analysis, and differences with $p < 0.05$ were considered statistically significant. The results obtained were compared with results reported in the literature.

Results

Subjects

After a review of more than 300 charts, a total of 114 patients were deemed suitable for inclusion. Certain patients were excluded because of incomplete data. Others were excluded if any stage of the cleft repair had been performed by surgeons other than the specified surgeon or his resident. Patients whose follow-up was inadequate and those who died before proper speech assessments could be completed were also excluded.

Of the 114 patients, 28 (25%) required secondary pharyngoplasty.

Sex

The patient pool consisted of 63 boys and 51 girls. Sex was statistically significant in determining the frequency of secondary pharyngoplasty; 21 (33%) of the 63 boys but

only 7 (14%) of the 51 girls required subsequent creation of a pharyngeal flap ($p = 0.025$).

Type of Cleft

The clefts were classified according to Veau's system,[2,3] which is not too complicated or too extensive and which is based on an anatomic landmark (the incisive foramen). Veau's system encompassed 4 categories: bilateral cleft lip and palate, unilateral cleft lip and palate, hard and soft cleft palate, and soft cleft palate. For this study, a fifth category was added, submucous cleft palate. Of the 114 patients in the study, 12 had bilateral cleft lip and palate, 38 had unilateral cleft lip and palate, 16 had hard and soft cleft palate, 33 had soft cleft palate, and 15 had submucous cleft palate.

Type of cleft was also significant in determining the need for subsequent pharyngoplasty ($p = 0.030$; Fig. 1). Patients with bilateral cleft lip and palate (6/12 or 50%) and hard and soft cleft palates (7/16 or 44%) were far more likely to require secondary pharyngoplasty than those with unilateral cleft lip and palate (8/38 or 21%), submucous cleft palate (3/15 or 20%) or soft cleft palate (4/33 or 12%).

Surgical Technique

Various techniques were used for initial closure of the cleft. In all patients, a 1-stage palatal closure was performed, in which the hard and soft palates were closed simultaneously. In these cases, the surgeon used a 3-layer closure, in which intravelar veloplasty was performed to reorient the soft palate muscles. For the 114 patients, 3 different 1-stage palatoplasty techniques were used: 79 patients underwent Veau-Wardill-Kilner type V-Y push-back operations,[4] 34 patients underwent von Langenbeck repairs,[4] and 1 patient underwent a Furlow z-plasty.[4]

Surgical technique was not a significant factor ($p = 0.35$) in determining subsequent pharyngoplasty. The single case involving Furlow z-plasty was excluded from the evaluation, although a secondary pharyngeal flap was required in that patient. The proportion of patients who needed secondary pharyngoplasty was the same (24%) for those who underwent the V-Y pushback procedure (19/79) and the von Langenbeck procedure (8/34).

Age at Primary Repair

The data were also analyzed according to age at primary repair. The patients were divided into clinically relevant groups with a sufficient number of patients in each group for statistical analysis. However, extreme values (older ages) were eliminated to prevent undue influence on the analysis, since the success of operations at older ages (beyond 18 months) may be influenced by factors other than the age at repair. A total of 97 patients underwent primary repair between 6 and 18 months: 24 patients between 6 and 9 months, 27 patients between >9 and 12 months, 26 patients between >12 and 14 months, and 20 patients between >14 and 18 months.

The age at primary repair was not significant ($p = 0.29$). Pharyngoplasty was most common in patients who underwent primary repair at age >9–12 months and least common in those first treated at age >12–14 months (Fig. 2).

Discussion

The results of this study correspond well with those reported previously. Twenty-five percent of the 114 patients required secondary pharyngoplasty following cleft repair. This value is similar to the mean of previously published reports (about 27%): 15% reported by Marrinan and others,[5] 45% by Mackay and others,[6] 30% by Dreyer and Trier,[7] 20% by Park and others[8] and 23% by Hartel and others.[9]

Sex was a significant factor in determining need for subsequent pharyngoplasty, but no literature sources addressing this subject were found. This observation raises several questions: Do the different growth patterns seen in boys and girls alter the outcome of the primary surgery? Should boys undergo primary repair at older ages than girls? A multivariate analysis might have been useful for answering these questions, but was limited by insufficient patient subgroup sizes.

Type of cleft was also a significant factor in determining need for pharyngoplasty. Clefts with unattached vomer were far more likely to require a pharyngeal flap. The vomer may therefore be a crucial feature. Perhaps cases in which the vomer is attached maintain more palatal length, which enhances the chance of effective closure during primary repair. Conversely, those with an unattached vomer may have hypoplastic musculature. The results reported here conform with this hypothesis: patients in whom the vomer was unattached (bilateral cleft lip and palate and hard and soft cleft palate) were statistically more likely to need secondary pharyngoplasty than those with attached vomer (soft cleft palate and unilateral cleft lip and palate). The results of Marrinan and others[5] support this idea; they also observed a difference in rate of creation of pharyngeal flaps between patients with attached vomer (unilaterial cleft lip and palate and soft cleft palate; 10%) and those with unattached vomer (bilateral cleft lip and palate and hard and soft cleft palate; 23%). These authors included submucous cleft palate with soft cleft palate. Park and others[8] reported that patients with bilateral cleft lip and palate were the most likely to require a pharyngeal flap (35%), although they did not differentiate hard and soft cleft palate from soft cleft palate. Krause and others[10] have also shown a relationship between the extent of the cleft (according to a gradient from soft cleft palate through hard and soft cleft palate and unilateral cleft lip and palate to bilateral cleft lip

and palate) and the outcome, with the most extensive defect being the most likely to require a pharyngoplasty.

In this study, it was felt that the data for patients with a submucous cleft palate might have interfered with the analysis, as these defects are often asymptomatic — that is, affected patients may have a competent velopharyngeal mechanism or the problem goes unnoticed until later in life. Therefore, the data were re-analyzed without data for patients with submucous cleft palate, but the same trends were observed for all analyses done on sex, type of cleft, technique and age at repair.

Technique was not a significant factor, as the V-Y push-back and von Langenbeck procedures rendered similar results. Marrinan and others [5] and Lin and others[11] also found no significant difference between surgical techniques. Optimal technique for primary repair continues to be widely debated. In this context, it is interesting to consider this patient group in a different sense. The initial chart review included 33 patients (beyond the 114 analyzed in this article) who underwent pharyngoplasty at the time of initial repair because the surgeon judged that the extremely wide defects could not be closed by simple mucoperiosteal flaps alone. These patients were excluded from the current analysis, but it might be instructive to consider their surgery as failed 1-stage repair (i.e., 1-stage repair alone was insufficient to yield normal velopharyngeal function). If these 33 patients were added to the patient pool, the frequency of pharyngoplasty would increase from 28/114 (25%) to 61/147 (41%). Although the result obtained when the 33 additional patients are included is not as similar to the values previously reported in the literature, perhaps it reflects a better way of analyzing the data. Perhaps the literature comparing 1-stage and 2-stage cleft repairs overlooks this important aspect, as some other centres[11] have reported pharyngoplasty at time of palatal repair. The question arises as to whether it might have been possible in these 33 patients to manage the defect without a pharyngeal flap if the soft palate had been repaired at a younger age.

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Another approach to closure of the cleft palate is the 2-stage repair, which has become quite popular in recent years. Jean Delaire and his colleagues, from Nantes, France, have developed a very logical 2-stage approach to cleft lip and palate, which is preferable in all cases of complete cleft palate, with or without cleft lip.[12–15] Delaire advocates simultaneous closure of the soft palate and the lip at 6 months of age. If the lip is not cleft, the soft palate is repaired at 9 months. The residual cleft of the hard palate is then closed at the age of 15–18 months, by which time, with rare exceptions, the cleft has become sufficiently narrow to be closed without or with only minimal displacement of the palatal fibromucosa. The theory is that closure of the soft palate allows for competence of the velopharyngeal sphincter to aid in speech development, while delayed closure of the hard palate reduces the effects on facial growth. Delaire feels that medial displacement of the posterior bony structures, the medial pterygoid plates and the maxillary tuberosities, continued growth of the palatal shelves, and further improvement of the relationship between the anterior parts of the bony segments occur more harmoniously with a 2-stage repair.

There are no studies comparing the Delaire approach to a 1-stage repair, but such research would be of interest. Other 2-stage repairs have been evaluated. Schweckendiek and Doz[16] and the Zurich group (Van Demark and others[17]) earlier described success with a 2-stage repair, although they delayed hard palatal closure until 5–12 years. The Zurich group found velopharyngeal insufficiency in only 5.4% of the 37 patients treated. They delayed hard palate closure until 5 years of age, which today is deemed too late for the repair. However, they felt that the same results could be achieved with earlier repair of the hard palate. Data from Rohrich and others[18,19] suggest that delaying hard palate closure beyond 18 months results in significant speech impairments without any beneficial response in maxillofacial growth. Vedung[20] found no difference between 2-stage and 1-stage closure. Of 328 patients who underwent 1-stage repair, only 6.7% required a pharyngeal flap and of 192 patients who underwent 2-stage repair, only 7.0% required pharyngoplasty. Hartel and others[9] obtained similar results in their follow-up study of 474 patients.

Age at repair was not a significant factor in this study. Other researchers have found significant differences with age at repair. Marrinan and others[5] found that the earlier a cleft was repaired (8–10 months was the youngest group), the less likely that patients would need a pharyngeal flap. Also, the need for a pharyngeal flap increased with age at primary repair (need for pharyngoplasty was greater for patients who were older than 16 months at primary repair than for those first treated at age 14–16 months, which was in turn greater than for those first treated at age 11–13 months). However, Ysunza and others[21] found no significant difference in velopharyngeal insufficiency between patients undergoing primary repair at 6 months and those undergoing primary repair at 12 months. The debate continues regarding the optimal timing of repair. Treatment protocols must balance the beneficial effects of

early repair on speech with the potential risk of impairment of maxillary or facial growth because of early repair.[1,22] The current trend seems to be toward early repair, between 6 and 12 months,[21] whereas others still feel that 12–18 months is the ideal time for cleft repair.[23,24]

This study had some limitations. The follow-up period was limited to 6 years in the later cases. The ideal study would be a long-term investigation following patients from birth until 10 to 15 years of age. In addition, the size of the patient pool (114) limited the statistical analyses. Some multivariate analyses were performed, but many of the subgroups in these analyses had to be excluded because of small sample size. A larger patient pool would have allowed more detailed statistical analyses, including more multivariant analysis. Other variables could have been analyzed, such as severity of the cleft, which can be measured from study casts, if available. Another potential variable would have been the surgeon's experience. Unfortunately, the charts reviewed in this study did not indicate whether the resident or the supervising surgeon performed the surgery. The patients' socioeconomic status might also be an interesting variable. For example, does socioeconomic status affect how much families do to help their children develop correct speech habits? Would such differences affect overall speech development? All of these factors should be analyzed in a properly controlled study.

Conclusions

In this study the frequency of velopharyngeal insufficiency after 1-stage palatoplasty was consistent with reported results. Sex and type of cleft were significant factors in the outcome of primary palatal closure. Of interest would be a study of primary palate repair at a young age comparing 1-stage and 2-stage approaches. Anatomically and theoretically, the 2-stage approach seems more appropriate for repairing the palate to normal function without affecting growth. ✦

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