Cleft Oronasal Fistula: A Review of Treatment Results and A Surgical Management Algorithm Proposal

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**Background:** Oronasal fistulas (ONF) following cleft palate repair are commonly encountered and remain a challenging problem. With reported recurrence rates between 33% and 37%, this urges us to critically evaluate the current treatment and propose a surgical management protocol.

**Methods:** A retrospective study of patients treated for ONF by a single surgeon between 1995 and 2005 was performed. Data regarding cleft type, age at palate repair, complications, location and size of fistula, tissue condition, surgical technique employed, and success rate were gathered.

**Results:** There were 64 patients (33 male and 31 female), and 44% of them had bilateral cleft lip and palate. Hypernasality and regurgitation were the major presenting symptoms of these patients with ONF. Fistulas mostly occurred in the hard palate area (53.1%). Severe scarring surrounding the ONF was reported in 31.2% of patients. Local flap and two-flap palatoplasty were the most common techniques (62.5%) used for closure of the ONF. Twenty-five percent of patients needed more than one repair to close the fistula. However, the overall success rate of closure was high (90.5%). Velopharyngeal (VP) function was significantly improved: only 26.8% of patients had adequate VP function before ONF closure and 64.3% patients had adequate VP function after ONF closure. However, the VP function of twenty patients remained inadequate or marginal.

**Conclusions:** A high success rate was achieved for closure of cleft ONF, although a certain percentage of patients required re-operation. Multiple fistulas and severely scarred palates made closure difficult. Successful closure of a fistula improved VP function but VP surgery was still indicated in certain patients. Based on the findings, an algorithm for management of cleft ONF was proposed.


**Key words:** oronasal fistula, cleft palate, outcome assessment, velopharyngeal function

The goals of palatal surgery are closure of the communication between the oral and nasal cavities, and construction of a functional velum that allows good speech production. Many techniques...
have evolved to achieve the primary goals of palate repair: single operation and primary healing. However, in some cases oronasal fistula (ONF) can be found after palate repair.(1) Cleft palate fistula is defined as a failure of healing or a breakdown in the primary surgical repair of the palate. Intentionally unrepaired nasoalveolar and/or anterior hard palate fistulas are not included in the condition.(2,3)

ONF following palate repair is not uncommon. The incidence varies greatly among centers and surgeons, and has been reported to be between 5% and 29%.(1) ONF has been associated with severity and type of cleft, repair technique, timing of repair and the experience level of the operating surgeon.(4,5) Once it occurs, ONF can be troublesome, not only for the patients but also for the surgeons. With the reported overall success rate of ONF repair approaching 85% and a recurrence rate of 33% to 37%, it remains a challenging problem.(1)

Many surgeons have proposed a variety of surgical methods for closure of ONF, from local flap to microsurgical repair. However, there have been few treatment protocols available to properly treat this condition. We reviewed all the patients treated for ONF in our center by a single surgeon and proposed a surgical treatment protocol.

**METHODS**

We retroactively reviewed 64 consecutive patients who underwent fistula closure performed by a single surgeon (LJL) at the Craniofacial Center, Chang Gung Memorial Hospital between July 1995 and July 2005. Non-surgically treated patients were not included in this study. All clinical data were gathered including gender, type of cleft, age at palate repair, symptoms, size, location, type of fistula repair, velopharyngeal (VP) function and outcome. Severity of palatal tissue scarring was also obtained. Severely scarred tissue was defined as multiple irregular scars on the palate with hard mucosal consistency and dense fibrotic tissue surrounding the fistula. VP function was evaluated by speech pathologists beginning at the age of 2.5 years. Further VP examination using nasoendoscopy and/or videofluoroscopy was performed at or after the age of 4 years if there were signs of hypernasality or nasal emission. VP function was determined using a combined clinical rating and the above examinations (Table 1).(6) Outcomes were measured by successful closure of the fistula, remaining symptoms and speech function.

<table>
<thead>
<tr>
<th>Table 1. Combined Velopharyngeal Competence Rating: Perceptual Speech, Videofluoroscopy and Nasoendoscopy</th>
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<tr>
<td><strong>Velopharyngeal competence</strong></td>
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<tr>
<td>Adequate</td>
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<tr>
<td>Probably adequate</td>
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<tr>
<td>Marginal</td>
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<td>Probably inadequate</td>
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**Abbreviations:** PPW: posterior pharyngeal wall; LPW: lateral pharyngeal wall.
RESULTS

This study group included 33 male patients and 31 female patients, with a mean age at fistula repair of 15.7 years (standard deviation (SD) = 11.7 years). The mean follow-up time after repair was 42.1 months.

General information

Bilateral cleft lip and palate (BCLP) was the major type, found in 28 (44%) patients. Twenty-six (41%) patients had unilateral cleft lip and palate (UCLP), 8 (13%) patients had cleft palate and two (3%) patients had submucous cleft. Most of the clefts were non-syndromic: 9 patients had other associated anomalies, including Pierre Robin syndrome, hemi-facial microsomia, Van der Woude syndrome, constriction bands and median facial dysplasia.

Thirty-four patients underwent initial palate repair in our hospital, while 46.9% (30 patients) of cases were referred from other hospitals. Since many of the cases were treated elsewhere, the age at initial palate repair was not available in these cases. From the available data regarding the initial palate repair, we found that almost the same number of patients were treated before 2 years of age (20 patients) as those who were operated on after 2 years of age (18 patients). From the latter group, 8 were operated on after the age of 10 years. Regarding the surgical methods for initial palate repair, 18 patients received the two-flap method, while 10 underwent Widmaier palatoplasty, 3 Sommerland technique, 2 Langenbeck technique and 1 push-back palatoplasty. The initial palate repair methods used in other hospitals were not clear from the chart records. Of the available data, we found wound dehiscence was the major post-palatoplasty complication. It was found in 14 patients. Two patients had flap necrosis and bleeding, while infection and injured pedicle were found in one case each.

Fistula signs and symptoms

Most of the fistulas were found in the first two weeks after palate repair, while in 6 cases they were found between 2 to 4 weeks after surgery. They were also noticed months after surgery due to loss to follow-up. Hypernasality and regurgitation were the major presenting symptoms, accounted for 39 and 28 patients, respectively.

The fistulas were mostly located in the hard palate (34 cases), followed by the junctional area of the hard and soft palate (23 cases) and 2 in the soft palate. Multiple fistulas of the hard and soft palate, and hard and junctional area were found in 3 and 2 cases, respectively. Of the 39 cases with available measurements of fistula size, we found that only 10% of patients had small fistulas of 1 to 2 mm, 36% of patients had medium-sized fistulas of 2 to 5 mm, and the majority of patients (54%) had large fistulas of more than 5 mm. Of the large fistulas, the size was between 5 and 10 mm in 12 patients (31%), 10 to 20 mm in 3 patients (8%) and more than 20 mm in 6 patients (15%). Severely scarred tissue, as defined by multiple irregular scars on the palate with hard mucosal consistency and dense fibrotic tissue surrounding the fistula, were reported in 20 patients (31.3%).

Surgical repair for ONF

Most of the patients underwent ONF closure after the age of four years, while in 21 patients ONF closure was performed between the ages 4 and 16 years, and in 31 patients it was performed when the patient was older than 16 years. Twelve patients underwent ONF closure before the age of 4 years. Irrespective of the surgical technique, we closed the palate fistulas with a two layer closure. Two-flap palatoplasty was the most chosen technique, being used in 24 patients (37.5%). Sixteen were closed with local flaps (25.0%), 5 with Langenbeck technique (7.8%), 6 with Furlow palatoplasty (9.4%) and 13 others required a tongue flap to close the fistulas (20.3%).

The majority of patients only needed one surgery to repair the fistula but, in 15 (23%) patients, a second surgery was required, and 3 patients required a third procedure. One patient refused further surgery. Six patients who needed repeated repair had a severely scarred palate. Bleeding and dehiscence after fistula closure were also found in one case each.

Of these failed cases, ten of them had no data of initial palatoplasty technique due to referral from another hospital. Two-flap technique and Widmaier palatoplasty were used in two cases each, while Sommerland and push-back technique were used in one case each. In these 16 failed cases, 7 had two-
flap palatoplasty as the chosen technique to close the fistula, 4 had a local flap, 4 had a tongue flap and one underwent Langenbeck technique. To overcome these recurrences, during the second operation, the surgeon chose a two-flap palatoplasty in 6 cases, a local flap in 5 cases, a tongue flap in 2 cases, and Furlow palatoplasty and Langenbeck method in one case each. However, recurrence of fistulas still occurred in 6 cases. Among these, only 3 patients underwent a third surgery, and were treated with a local flap, a two-flap palatoplasty and a tongue flap (Table 2).

**VP function**

VP function was reviewed in 56 patients and revealed that 73.2% of patients had inadequate or marginal VP function before ONF closure. Only 15 patients (26.8%) had adequate function. Most of the patients with inadequate or marginal function had fistulas that were located in the hard palate and junctional areas. Large fistulas were found in 63% of patients with VP dysfunction. However, small fistulas were also observed to cause the same problem.

After ONF closure, VP function was adequate in 64.3%, with two patients requiring additional VP surgery. The improvement in VP function was significant (Figure 1). However, it should be noted that twenty patients continued to have VP insufficiency, either marginal or inadequate.

**Outcome**

Although repeat operations were needed in 25% of patients for closure of the fistula, overall successful closure was achieved in 90.5% of patients. From the chart review in this study, we also found that these ONF patients had quite a high incidence (53.1%) of maxillary hypoplasia.

**Table 2. Surgical Oronasal Fistula Closure Methods**

<table>
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<tr>
<th></th>
<th>1st operation</th>
<th>2nd operation</th>
<th>3rd operation</th>
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<tbody>
<tr>
<td>Local flap</td>
<td>16</td>
<td>5</td>
<td>1</td>
</tr>
<tr>
<td>Langenbeck palatoplasty</td>
<td>5</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>Two-flap palatoplasty</td>
<td>24</td>
<td>6</td>
<td>1</td>
</tr>
<tr>
<td>Furlow palatoplasty</td>
<td>6</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>Tongue flap</td>
<td>13</td>
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**DISCUSSION**

We retrospectively studied 64 patients treated for ONF during a 10 year period. This study is valuable since all of the cases were operated on by a single experienced surgeon, and had complete and long-term follow-up of 42.1 months in one institution. The fistula recurrence rate of 25% was lower than other reported series of 33% to 37%. Furthermore, the overall success rate of 90.5% was higher when compared to other series, which usually had rates of less than 85%. Therefore, thorough evaluations were performed, especially regarding the surgical technique applied, to make some recommendations for choosing which surgical technique should be employed.

ONF has been reported to be associated with severity and type of cleft. From our series, we found that BCLP was the major type of cleft, similar to other reported series, and UCLP were also comparable to this group. Only a very few patients had cleft palate or submucous cleft. Complication by a palatal fistula represents a technical failure resulting from poor wound healing, tension or absence of multilayer repair. In our series, we found it was mostly caused by wound dehiscence. Other causes are bleeding, partial mucoperiosteal flap necrosis, infection and injury of the pedicle. Most of the fistulas were observed in the early stages, within two weeks after palatal surgery.

This condition has functional consequences because of fluid and air leakage. Air leakage may cause speech impairments due to nasal escape. Even
very small fistulas, measuring 1 to 2 mm, can cause VP disturbance. Leakage of fluids and lodging of food particles can be troublesome for patients. Most of our patients came with speech-related problems such as hypernasality, nasal emission and VP insufficiency. Regurgitation was also a major presenting symptom.

It is important to perform a thorough examination of the fistula. A simple illumination test using a nasopharyngoscope can allow clear inspection of the fistula. The nasopharyngoscope also helps to evaluate soft palate mobility and the competency of the VP closure. To examine possible influences during speech, the fistula should be temporarily covered, using dental wax or a palatal plate. Speech quality is examined before and after coverage. If nasalization diminishes after covering, the nasality is mainly caused by the fistula. A period of speech therapy may be needed to unlearn VP dysfunction. On the other hand, if the nasalization persists, an additional VP surgery should be considered. This simple method makes decision-making more effective.

The critical limit of fistula size that has an adverse effect on speech has been widely discussed. Many suggest that an area exceeding 4.5 mm² to 5 mm² could interfere with speech. It usually causes hypernasality, audible nasal escape and weakness of pressure consonants. However, some authors have found that a fistula of only a few millimeters square can affect speech and resonance. This is similar to our findings: despite the fact that the majority of our patients had VP dysfunction related to fistulas measuring more than 5 mm, a small fistula could also cause VP dysfunction.

In the early stages after the primary palate repair, if an ONF occurred, we treated the fistula conservatively. In most cases the fistula will spontaneously narrow or even close, in the case of small fistulas. Conservative treatment is also indicated for an asymptomatic fistula. The indications for fistula repair relate to the associated symptoms, as described previously. It has been reported that fistulas causing disturbance in speech should undergo early repair, whereas the closure of fistulas not associated with speech problems should be delayed, if possible, until completion of orthodontic maxillary arch expansion and be combined with secondary alveolar bone grafting. It has been reported that orthodontic treatment can cause disruption of the repaired fistula. However, the psychological and social impact of the symptomatic ONF warrants early surgical treatment. If indicated, an early closure at any age is our most preferred option. This can be achieved after thorough discussion about the complications with the parents. Possible ONF recurrence during palatal expansion can be managed later. For management of an ONF associated with VP insufficiency, it is preferred to wait until 4 years of age when an objective nasopharyngoscope or videofluoroscope examination can be performed.

Surgical management is a popular and effective method to close ONFs. The principle of surgery is to perform two layer, tension free closures in all cases. Many surgical techniques have been employed to fulfill this objective, from local flap to free tissue transfer. This may reflect the difficulty in treating this condition. From the literature it is known that a surgeon can make a regional flap by utilizing adjacent tissue, depending on the location. A turn over flap and transposition flap from buccal, vestibular or facial regions can be used, along with tissue from the nasal cavity, septal or turbinate. However, it should be kept in mind that these can cause growth disturbance, donor site morbidity or visible scars. It is our preference to use local palatal mucosa or mucoperiosteal tissue to replace the defect, based on the principle of replacing tissue with like tissue.

For large fistulas, where it is judged impossible or difficult to use local tissue, a tongue flap may be employed. A distally based, midline positioned tongue flap with the ratio 1 to 1.5 up to 5 can be used without causing donor site problems or flap circulatory disturbance. With this type of tongue flap, the donor scar is acceptable, leaving a fine linear scar in the middle of the tongue, the possibility of injuring the taste buds is avoided and there is no interference with tongue mobility. This flap is beneficial for recurrent, recalcitrant fistulas with extensive scarred tissue. Therefore, a careful examination of the condition of the tissue, especially concerning scarring, should be properly performed. With an extremely large fistula, heavily scarred palate and contracted dentoalveolar arch, orthodontic palatal expansion may be required followed by a free tissue transfer. However, this kind of case is rare. Several donor sites are available for free tissue transfer to close a huge ONF, such as the scapula, iliac crest, radial forearm and dorsalis pedis flaps. A radial forearm
flap is preferred since it is thin, pliable, variable, mostly hairless and has a long vascular pedicle.\(^{(15-17)}\)

For a wide and long ONF in the hard palate or multiple fistulas, a redo palatoplasty, i.e. two-flap palatoplasty, is carried out to achieve a two layer, tension free closure. In our series, a two-flap palatoplasty technique was frequently used based on this fact. The success rate was relatively high when compared to other series.

Palatal ONFs may require surgery if there is loss of intraoral pressure during speech. Correction of weak pressure by closing the fistula may facilitate speech. Medium-sized and large fistulas in this area require a relaxing incision in addition to a two layer closure. The goal of fistula correction in the soft palate is not only complete closure of the defect but also achieving a functioning soft palate that has adequate VP closure. For this purpose, it is better to perform a reoperation using a two-flap palatoplasty. This will close the fistula and improve the VP function mechanism during speech. A Furlow palatoplasty with double opposing z-plasty can be performed for an ONF in the hard and soft palate junctional area. It has the benefits of closing the fistula, reducing the wound tension in the fistula area and also lengthening the soft palate. The latter advantage is important since patients with an ONF are at high risk of speech deterioration at around the age of 9 years due to maxillofacial growth and involution of the adenoid tissue. With the soft palate lengthening effect obtained with the Furlow palatoplasty technique, VP insufficiency is prevented and, thus, further VP surgery. In the Chang Gung Craniofacial Center, Furlow palatoplasty is effectively used for treatment of marginal VP insufficiency.\(^{(18)}\)

In our series, there were a number of patients with large fistulas. However, most of the cases could be closed with a two-flap palatoplasty technique or tongue flaps. No patients were treated using free tissue transfer. For patients with large or very large fistulas and severely scarred palatal tissue, a tongue flap or microsurgical free flap transfer is recommended. Speech evaluation can be performed postoperatively to determine the need and type of further VP surgery. In summary, regarding selection of surgical procedures for closure of fistulas, a local flap is used for closure of small fistulas, a two-flap or Langenbeck method for small to medium-sized fistulas, the Furlow method for fistulas over the soft palate associated with VP insufficiency, and a tongue flap or distant flap for large or difficult fistulas. Factors that may influence selection should be carefully evaluated, such as scar condition, location of fistula, condition of the patient and the surgeon’s familiarity with the surgical methods. These principles are also applied for selection of second or third fistula closure attempts. In this palatal surgery category, the same or a different surgical method can be used on the same patient.

Significant improvement in VP function was noted in this study. After ONF closure, VP function was adequate in 64.3% of patients and two patients needed additional VP surgery. Twenty patients continued to have VP insufficiency, either marginal or inadequate. This finding suggests that successful closure of a fistula can improve VP function but VP surgery is still indicated in certain patients, indicating incomplete management. By performing a fistula occlusion test and nasopharyngoscope examination before surgery, these patients can be treated with simultaneous ONF closure and a VP surgical procedure. The fistula occlusion test can be performed simply by using a piece of chewing gum or dental palatal plate.

Based on these findings, an algorithm for management of cleft ONF is proposed (Figure 2). As a summary of our protocol, observation is suggested of non-symptomatic patients. For symptomatic patients, a perceptual VP assessment is performed. If VP function is normal, ONF closure can be performed at any age. For patients with inadequate VP function, nasopharyngoscope or videofluoroscope examination and a fistula occlusion test are performed at 4 years of age. If there is improvement, an ONF closure alone can be performed. Surgical technique is selected depending on the condition of the fistula. If the fistula is small, with mild scarring of the surrounding tissue, a local flap can be safely used. For medium-sized or large fistulas, a redo palatoplasty is recommended. This may also be combined with a Furlow palatoplasty. Furlow palatoplasty is useful for treating fistulas in the junctional area. This is a nice procedure with less patient discomfort, and the advantages of avoiding tension in the fistula area, lengthening the soft palate and, thus, being beneficial in avoiding future possible VP deterioration. For patients with no improvement shown on the fistula occlusion test, a VP surgical procedure is performed...
simultaneously with an ONF closure. This can be a Furlow palatoplasty for patients with marginal VP function, or a pharyngeal flap or sphincter pharyngoplasty for patients with inadequate VP function. In our center, a pharyngeal flap is preferred due to the lower rate of nasal airway obstruction when compared to a sphincter pharyngoplasty. For patients with very large ONFs and severe scarring of the palatal tissue, a tongue flap or, rarely, free tissue transfer is recommended. In this case, VP surgery should be postponed to a later stage to avoid the risk of airway obstruction. In this group of patients, the VP function usually remains inadequate despite some improvement after fistula closure.

REFERENCES

3. Muzaffar AR, Byrd HS, Rohrich RJ, Johns DF, LeBlanc D, Beran SJ, Anderson C, Papaioannou A. Incidence of...
唇顳裂口鼻癢管：治療結果的回顧和手術計畫的提議

Enrina Diah 羅綸洲 雲天湘 王淑慧 Luh K. Wahyuni 陳昱瑞

背 景： 頭裂手術後發生的口鼻癢管，是一項常見而充滿挑戰性的問題。文獻報告其發生率在33%至37%之間，所以有必要檢測目前的治療狀況，研擬適當的手術計畫。

方 法： 這項回顧性研究，收集一位醫師在1995至2005年間，所治療過口鼻癢管的病人。資料收集包含唇顳裂型態、顳裂手術年齡、併發症、癢管的位置及大小、顳組織情況、手術方法，以及成功率。

結 果： 計有33位男性，31位女性，共64位病人，其中44%為雙側唇顳裂。最常見症狀是鼻音過重和逆流，癢管在硬顳部位最多，佔53.1%。31.2%的病人癢管周圍組織的疤痕化嚴重。局部皮瓣和兩瓣式頜修補術是最常用的方法，佔62.5%。25%的病人，需要二次以上的手術來關閉瘜管，然而總體的成功率高達90.5%。顳咽功能則有顯著的改善，從術前的26.8%適當率，手術後升高至64.3%，然而仍有20位病人的顳咽功能沒有達到正常。

結 論： 雖然有些病人需要一次以上的手術，口鼻癢管的手術閉合成功率很高。多發性癢管和疤痕嚴重者，治療比較困難。癢管手術閉合成功者，顳咽功能會有改善，然而對於某些病人，顳咽手術還是需要的。根據這些研究成果和證據，吾人提出唇顳裂口鼻癢管的治療流程。
（長庚醫誌2007;30:529-37）

關鍵詞：口鼻癢管，顳裂，結果評估，顳咽功能