Reconstruction of Large Anterior Palatal Fistulae Using Tongue Flaps

Geniş Anterior Palatal Fistüllerin Dil Flepleri İle Rekonstrüksiyonu

INTRODUCTION
Palatal fistulas represent an important concern in the treatment of cleft palates (1). The first reference to the use of the tongue to close palatal fistulae after cleft palate repair is the paper by Guerrero-Santos and Altamirano (1966) (2). Anatomically, the tongue has a rich vascular supply. This is predominantly from the lingual artery which branches into the dorsal lingual, deep lingual and sublingual arteries. It also has a secondary supply from the tonsillar branch of the facial artery and the ascending pharyngeal artery. Consequently, the blood supply is abundant with a rich submucous plexus of vessels on the dorsum of the tongue that provides an excellent site to raise either a dorsal or anterior-based flap (3). There are many methods proposed for closure of palatal defects.

Large defects present a more difficult problem. The aim of this paper is to report our experience with a large amount of tissue that they provide has made tongue flaps particularly appropriate for the repair of large fistulas in palates scarred by previous surgery. We recommend this as a reliable surgical technique for the closure of palatal fistulas.

Naouar Ibnouelghazi¹, Zakaria Aziz², Jinane Kharbouch¹, Salma Aboulouidad³, Nadia Mansouri-Hattab¹

¹Oral, Maxillo-facial and Aesthetic Surgery Department, Mohammed VI University Hospital, Marrakesh School of Medicine, Cadi Ayyad University, Morocco

Address correspondence to: Naouar Ibnouelghazi, Oral, Maxillo-facial and Aesthetic Surgery Department, Mohammed VI University Hospital, Marrakesh School of Medicine, Cadi Ayyad University, Morocco e-mail: ibnounawar@gmail.com

ORCID
Naouar Ibnouelghazi
https://orcid.org/0000-0002-7250-9127
Zakaria Aziz
https://orcid.org/0000-0001-5382-2372
Jinane Kharbouch
https://orcid.org/0000-0002-7871-8755
Salma Aboulouidad
https://orcid.org/0000-0002-4588-1353
Nadia Mansouri-Hattab
https://orcid.org/0000-0002-5764-2851

Abstract
Aim: The tongue is a most suitable donor site for the closure of cleft palate fistulae. The aim of this prospective study was to reassess the efficacy of tongue flap technique.

Patients and Methods: 4 cleft patients who underwent reconstruction by anteriorly based tongue flap between 2015 and 2017 were studied. Variables such as flap’s ability to close the fistula, the remaining tongue shape at least 1 year after surgery, and speech improvement (patient’s self-assessment and parents’ opinion) were evaluated.

Results: 4 cases presenting with type IV palatal fistulae and one type V were operated on using a double-layer closure with tongue flap. The average age at the time of the intervention was 68.5 years. The initial malformation was a complete unilateral cleft for 2 patients, and a palate velar cleft for the 2 others. The size of the fistulas was variable in length (7 to 12 mm). The procedure was successful as no recurrence of fistula was noticed at an average follow-up of 18 months, with full recovery of tongue esthetic and function.

Conclusion: Tongue flaps are used in cleft palate surgery because of their excellent vascularity, and the large amount of tissue that they provide has made tongue flaps particularly appropriate for the repair of large fistulas in palates scarred by previous surgery. We recommend this as a reliable surgical technique for the closure of palatal fistulas.

Key words: Myotonic dystrophy; cardiac manifestation; presyncope

Disclosure: None of the authors has a financial interest in any of the products, devices, or drugs mentioned in this article. The research was not sponsored by an outside organization. All authors have agreed to allow full access to the primary data and to allow the journal to review the data if requested.

the use of tongue flaps for the repair of large palatal defects in adult cleft patients. A number of fistulae can be closed with local tissue using turnover flaps based on a margin of the defect to provide nasal layer cover. Almost always these flaps must be based on a scar at the margin of the defect and their blood supply is, therefore, necessarily somewhat precarious.

Oral layer cover can often be provided by transposition flaps from local palatal tissue or, in the presence of a complete cleft, by buccal sulcus flaps introduced into anterior hard palate defects through the alveolar defect. However the introduction of a tongue flap with its bulk of vascular soft tissue now makes it possible to close fistulae previously considered inoperable. It is a last chance option. The indications for closing palatal fistulae include: The embarrassment of fluids leaking down the nose and to a lesser extent, rhinitis and catarrh produced by food lodging in the nose. Interference with articulation. This feature is not uncommon in those patients who develop a habit of closing the fistula with part of their tongue. In other patients audible nasal escape and hypernasal resonance can mar the quality and reduce the volume and continuity of speech.

We used the Pittsburgh Fistula Classification System includes seven fistula types (Figure. 1). Fistulas at the uvula, or bifid uvula, are considered a type I fistula. Type II fistulas occur within the soft palate. Type III fistulas are found at the junction of the soft and hard palates. Type IV fistulas are located within the hard palate. Type V fistulas are defined as fistulas at the incisive foramen (junction of the primary and secondary palates) in the context of Veau IV clefts. Type VI fistulas are lingual- alveolar, and type VII fistulas are labial-alveolar fistulas. In this study, we limited ourselves to fistulas located within the hard palate Type IV and the junction primary/secondary palate Type V.

PATIENTS AND METHODS

4 cleft patients who underwent reconstruction by anteriorly based tongue flap between 2015 and 2017 were studied. Variables such as flap's ability to close the fistula, the remaining tongue shape at least 1 year after surgery, and speech improvement (patient's self-assessment and parents' opinion) were evaluated.

The surgical procedure was as follows:

1. **First operational time**

1.1. Installation

The procedure was conducted under general anesthesia with nasotracheal intubation; the patient was placed in a dorsal position with the head resting on a headrest. After disinfection of the oral cavity with polyvidone iodine, the loss of substance from the palate is measured with a graduated ruler. The tongue was then maintained outside the oral cavity by suture in the apex of tongue, maintaining its symmetry, so that the incision traces could be drawn. Incision was performed around the fistulous tract to design a local flap for nasal closure; mucosalized edges were excised. The nasal layer was identified and carefully dissected then inversed margins are sutured without tension using 4-0 Vicryl.

1.2. Raising the anterior-based tongue flap

The flap is delineated with a marking pen. The cleft
length dictates its length, and the flap should sit passively. The width should be slightly greater than the cleft defect and the depth of the flap should be approximately 3 mm, to include intrinsic muscle, which may deepen to 5 mm toward the base. Up to 2/3 of the dorsal surface may be used without significant tongue deformity. Care must be taken not to extend the flap too close to the circumvallate papillae or the tip of the tongue. Adequate hemostasis is maintained throughout with bipolar diathermy. The donor site is closed with 4.0 resorbable suture taking care not to constrict the pedicle base. The flap is positioned within the defect. It is sutured into the reception site with a 4.0 resorbable suture, anteriorly and laterally. Nasogastric tube was placed to assist in feeding.

2. Second operational time

After 15 days to 21 days, the patient was taken back into the operation theater; under local anesthesia, the flap was divided and set into the posterior aspect of the palatal defect. The donor site defect was closed using 4-0 Vicryl.

Patients were assessed under the following criteria at 1 week, 1 month, 3 months, 6 months, and thereafter at 1-year intervals: (1) flap viability; (2) fistula closure; (3) residual tongue function and aesthetics; (4) assessment of speech impediment. Length of follow-up period ranged from 2 weeks to 18 months, with an average length of 15 month.

3. Postoperative care

Feeding is via a nasogastric tube between two operating times, to facilitate the cicatrization. Oral hygiene must be rigorous and include baths of mouth of an iodine solution. A few days after the second operative time, the nasogastric tube is removed. The food is then mixed until the palate is healed.

RESULTS

4 cases were operated on using a double-layer closure with tongue flap (Table 1). Most of the fistulas reviewed were of Type IV as per the Pittsburgh Fistula Classification System (1). The success rate of these 4 cases was 100%, with no recurrence of fistula at an average follow-up of 18 months. The study was prospective, on 4 patients. They had a type IV secondary palate fistula. The average age at the time of the intervention was 68.5 years. The initial malformation was a complete unilateral cleft for 2 patients, and a palate velar cleft for the 2 others. The size of the fistulas was variable in length (7 to 12 mm), but never exceeded 1 cm in width. The history of attempted closures ranged from 1 to 3. The procedure was well tolerated by our patients, and our outcomes demonstrate excellent healing both short and long term. None of our patients had premature flap dehiscence or necrosis and there were no complications at the tongue donor site. There have been no problems with tongue function postoperatively, and patients have reported no problems with feeding. There have been no palatal fistulae reported.

Speech was reported to have improved, but this will be assessed more formally. Table 2 shows patient’s satisfaction towards Tongue’s function and esthetic and speech improvement. The tongue flap is simple and reliable for the reconstruction of relatively extensive palatal defects. It has the advantage of preserving the function and aesthetics of the donor site and the recipient site. Primary treatment of cleft palate should result in an intact palate with separation of the oral and nasal cavities. However, the published reports of large series indicate fistula can recur in the secondary palate of a small but significant group of patients; the incidence varies from 8.9% to 34% (4). Fistulas may occur in the labial vestibule, the alveolus, the hard palate, and at the junction between the hard and soft palate. Symptoms of these fistulas may be hypernasality in speech, regurgitation of fluids into the nose, and food lodging in the defect. The symptoms depend to some extent on the site of the fistula.

In accordance with literature, most of the fistulas were located in anterior palate. In our study, The

Table 1. Demographic Data of the Patients

<table>
<thead>
<tr>
<th>Age (yr)</th>
<th>Gender</th>
<th>Number of Prior Closures</th>
<th>Size (cm)</th>
<th>Type</th>
<th>Flap Orientation</th>
<th>Follow-up</th>
<th>Complication</th>
</tr>
</thead>
<tbody>
<tr>
<td>Case 1</td>
<td>13</td>
<td>Male</td>
<td>2</td>
<td>10 mm</td>
<td>IV</td>
<td>Anteriorly based</td>
<td>1 year</td>
</tr>
<tr>
<td>Case 2</td>
<td>19</td>
<td>Female</td>
<td>3</td>
<td>7 mm</td>
<td>V</td>
<td>Anteriorly based</td>
<td>6 months</td>
</tr>
<tr>
<td>Case 3</td>
<td>26</td>
<td>Female</td>
<td>1</td>
<td>12 mm</td>
<td>IV</td>
<td>Anteriorly based</td>
<td>1 year</td>
</tr>
<tr>
<td>Case 4</td>
<td>42</td>
<td>Male</td>
<td>2</td>
<td>9 mm</td>
<td>IV</td>
<td>Anteriorly based</td>
<td>2 years</td>
</tr>
</tbody>
</table>
Pittsburgh Fistula Classification System was used to describe fistula. Most of the fistulas located within the hard palate Type IV (1). Attempts at closure using only local transposition flaps may be successful, although frequently this is not achieved and a smaller oronasal fistula will recur. Additional attempts to gain closure with local tissue alone often result in repeated failure as thick and immobile scarred palatal mucoperiosteum leads to closure under tension with subsequent flap necrosis and wound dehiscence. A variety of both surgical and prosthetic solutions to the problem of inadequate local tissue have been sought. The FAMM (facial artery musculomucosal) flap as introduced by Pribaz et al is a valuable option to reconstruct moderate-size defects of the anterior palate. The FAMM flap has few minor drawbacks; first, the pedicle may be injured during mastication to the point that the pedicle gets severed (5). And speech therapists discourage the use of a flap that, with the inclusion of facial muscles, will likely interfere with further speech development. Tongue flaps have been used to close intraoral defects following tumor surgery, severe infection, trauma, and cleft palate fistulas. According to De Santo, tongue flaps are also useful after radiation therapy. Posteriorly based flaps are indicated when treating defects of soft palate, retromolar region, floor of the mouth, and posterior buccal mucosa.

Anteriorly based flaps are useful in the treatment of defects of the hard palate, anterior buccal mucosa, lips, and anterior floor of the mouth. Pigott et al (2) presented their work on 20 patients with palatal fistulas, which were successfully closed with tongue flaps in 17 patients (85%). Contreras et al used tongue flap and forehead flap for closure of residual oronasal fistula, and they concluded that when all other surgical possibilities have failed, tongue, vestibular mucosa, pharynx, or forehead flap can be used to restore mucosal continuity (8). The authors concluded that the anteriorly based dorsal tongue flap is a safe and effective method for closure of relatively large palatal defects. The parameters for success include sufficient length of the flap (5 to 6 cm), a flap width somewhat larger than the defect, and a flap thickness of 0.5 cm (7). Alsalman et al. (15) used the single-layer closure for closing a palatal fistula.

Many surgeons have emphasized a double-layer closure for closing a palatal fistula to ensure non-recurrence (4, 9-11); some surgeons have introduced a 3-layer closure arguing better outcomes (12, 13) Some surgeons choose to limit the movement of the tongue to ensure better healing by fixing or anchoring the tongue flap to the nasal septum. Others choose more radical options like intermaxillary fixation or button suture to the lip, which makes the procedure more uncomfortable to the patients and limits the possibility of feeding (14). This was not done in any of our patients. Too much of the tongue movement is automatically restricted because of the pain

<table>
<thead>
<tr>
<th>Preservation of Tongue mobility function</th>
<th>Preservation of Tongue sensatory</th>
<th>Preservation of Tongue esthetic</th>
<th>Speech Improvement</th>
</tr>
</thead>
<tbody>
<tr>
<td>% Yes 100%</td>
<td>Yes 100%</td>
<td>Yes 100%</td>
<td>None 0%</td>
</tr>
<tr>
<td>% No 0%</td>
<td>No</td>
<td>No</td>
<td>Yes 100%</td>
</tr>
</tbody>
</table>

Table 2. Patients evaluation of speech improvement and tongue function

Figure 2. (Case 1). A. Preoperative palatal fistula; B. Fistula complete closure after 21 days postoperatively; C. Shape of tongue after tongue flap pedicle devision; D. Result 8 month after palatal fistula closure using anteriorly based tongue flap; E. Preservation of tongue esthetic at 8 months postoperatively
associated with it. Mahajan et al. (15) reported a series of 41 cases, where they used tongue flaps for closure of palatal fistula with no additional fixation of the flap. Many surgeons have emphasized a double-layer closure for closing a palatal fistula to ensure non-recurrence. (4, 9,10). Alsalman et al (16) used a modified single layer closure with tongue flap in cleft patients. In addition, some surgeons have introduced a 3-layer closure, arguing better outcomes. (12,13) In small fistulas, the nasal layer closure can be achieved by direct closure of the nasal layer or mobilizing some of the nasal mucosa.2,3

However, in large fistulas or excessively scarred palatal nasal layer, it is difficult to achieve closure in most cases. Therefore, a more robust flap, such as buccal flap, pharyngeal flap, or inferior turbinate flap, may be needed to close the nasal layer. We prefer to close the nasal layer using 2-layered reconstructions. Flap division has been done by various authors varying from 10 to 21 days (9, 17). In our series, we chose to do flap division after a period of 3-week. The tongue flap when compared to other reconstructive options has the advantage of an abundant vascular structure with significantly decreased rates of fistula recurrence after surgery. The second advantage of dorsal tongue flaps is the ease of planning the flap in sufficient length, width, and depth needed for the location and dimension of the fistula. The tongue flap has the advantage of giving a good volume by adding muscular tissue.

CONCLUSION

The Tongue flap is a reliable option to close a complicated palatal fistula. The study justifies the use of tongue flap for closure of palatal fistulas, as it provides abundant tissue with esthetic morbidity of the donor site. Hence, we would recommend this as a reliable surgical technique for the closure of palatal fistulas.

Conflict of interest: Authors declare that there is no conflict of interest between the authors of the article.

Financial conflict of interest: Authors declare that they did not receive any financial support in this study.

Address correspondence to: Naouar Ibnouelghazi, Oral, Maxillo-facial and Aesthetic Surgery Department, Mohammed VI University Hospital, Marrakesh School of Medicine, Cadi Ayyad University, Morocco e-mail: ibnounawar@gmail.com

REFERENCES