



CASE REPORT

Pediatric/Craniofacial

Double Tongue Flaps for Anterior Huge Palatal Fistula Closure

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Summary: Even though it is widely accepted that the tongue flap is effective and feasible for repair of huge palatal fistula, there still exist a few failed cases due to the severity or complicated situation. The aim of this paper is to report the validity and feasibility of using double tongue dorsal flaps to repair a huge anterior fistula. A 10-year-old boy diagnosed with Van de Woude syndrome with repaired bilateral cleft lip and palate presented with a huge anterior fistula divided by septum. A double tongue dorsal flap was designed to cover the fistula. The huge unusual anterior palatal fistula was repaired successfully by usage of double pedicle tongue flaps with a follow-up period of 1 month. The double tongue flap is an alternative choice to handle a large residual fistula in anterior part of palate which was divided into 2 fistulas by septum. This technique was indicated in the situation of large residual fistula in anterior part of palate which was divided into 2 fistulas by septum after bilateral cleft palate repair. (Plast Reconstr Surg Glob Open 2019;7:e2246; doi: 10.1097/GOX.000000000000002246; Published online 28 May 2019.)

alatal fistula is one of the most common complications for cleft palate repairing, and the incidence varies in great degree ranging from 3% to 60%1 with an overall incidence of 8.6%.2 The sizes and locations of fistula also differ from one another.3 A huge fistula in the anterior hard palate, which occurs more commonly in the case of bilateral cleft lip and palate, is always a big challenge for the surgeon. It has been well accepted that tongue flap is one of the methods that is mostly utilized to repair palate fistula, although several other operation procedures have also been used.4-6 Firstly introduced by Guerrero-Santos J and Altamirano JT in 1966,⁷ the protocol of the tongue flap has been described extensively in the literature.8-10 Nevertheless, there is no study demonstrating the application of double tongue flaps method in palate fistula repairing surgery as of today. In this paper, the authors described a new method—double dorsal tongue flaps—for the repair of huge anterior palate fistula.

PATIENT AND OPERATIVE PROCEDURE

A 10-year-old boy suffering from Van der Woude syndrome presented a huge fistula in the anterior hard palate

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with a surface area of approximately $4.5\times5\,\mathrm{cm}$, which was separated by the nasal septum (Fig. 1). The boy was born in September of 2008. The patient went through his first lip repairing surgery when he was 1 year old, and palate repairing when he was 7 years old. The wound in the anterior hard palate broke down 7 days after the surgery and the fistula was formed, which caused speech distortion and hypernasality when he spoke. Nasal regurgitation was also presented.

Operative Procedure

- 1. The operation was performed under general anesthesia with nasotracheal intubation.
- 2. Preparation of donor site: double bilateral dorsal tongue flap with anterior pedicle was designed, local anesthetic injection (1% lidocaine: adrenaline = 200,000:1) was given and 2 flaps (width around 2 cm × length around 5 cm) with full thickness of 3 mm were raised (Fig. 2) by using monopolar electrotome. Bleeding spots were carefully checked and stopped without sutures.
- 3. Preparation of recipient site: an incision was made around margins of the fistula slightly toward the oral side and both sides of vomer; mucoperiosteum with approximately 7mm in width was kept on the vomer bone. The mucosa of palatal margins and vomer flaps for both sides were mobilized and were closed with 4-0 Vicryl sutures to reconstruct the nasal layer.
- 4. Two tongue flaps were positioned to the recipient sites with suturing (1-0 silk). Nasogastric tube was placed and elastic band was used to limit mouth opening after the patient was extubated and conscious.

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Fig. 1. The fistula on anterior hard palate with deviated mobile premaxilla.

5. Buccal fistular closure: The cuttings were made around the labial buccal vestibular fistula on both sides and the mucous-periosteum membrane were lifted on labial and nasal side. The nasal layer was formed by suturing the nasal side mucous-periosteum membranes as the recipient site. The last step was to secure the tongue flap by suturing the mucous-periosteum membrane with the free end of the tongue flap.

RESULTS

Two pedicles were divided from tongue 2 weeks after the tongue flap operation. Firstly, 2 pedicles were divided



Fig. 2. Double tongue flaps (width around $2 \text{ cm} \times \text{length}$ around 5 cm) were lifted up with about 3 mm thickness.



Fig. 3. Picture of tongue dorsal view taken 1 month after the division of tongue flap pedicles.

under local anesthesia. Secondly, patient was intubated orally for residual tongue flaps in the donor side and the palatal side. Patient obtained complete healing and the results 1 month after the operation are shown in Figures 3 and 4.

DISCUSSION

Tongue flap procedure has been used for repairing soft tissue defects on lip and intraoral defects on cheek and oronasal communications caused by trauma or tumor resection for many years. 11,12 The fistula of the hard palate caused by previous attempts of cleft palate repairing was a major kind of unwanted oronasal communications, and it is a big challenge for the surgeon to repair it. In 1966, Jose Guerrero-Santos suggested the use of tongue flap to repair hard palate fistulas introduced by complications from cleft palate surgery, where 3 cases out of 10 were illustrated. Since then, many studies had been reported where the authors generally discussed the indications, procedures, tongue flap fixation, management of the complications, flap blood supply, and pedicle position. 9,10,13–25 Despite the fact that tongue flap has been proven to be a sturdy, reliable, and efficient means for the closure of hard palate fistula especially in the anterior region, there existed unsuccessful cases due to the over mobility of tongue, and sometimes the massive size of fistula. 4,10,26 For the unique case presented in this article, the anterior fistula due to its irregular shape and extended size of 4.5×5 cm was by far the largest fistula comparing with the ones reported in literature. In our case, a double anterior pedicle tongue flaps was designed for the patient based on the following reasons and principles: Firstly, tongue was divided into right and left halves by a medial fibrous septum. The vascular vessels of tongue were distributed on both sides of the midraphe. Poor blood supply in the midline of the tongue lowers the success rate of conventional tongue flap procedure, which required a midline cross to fully cover the large defect in the patient presented in this study, which may as well result in a secondary deformity



Fig. 4. Picture of palatal view taken 1 month after the division of tongue flap pedicles.

of tongue after suturing up the donor site. Secondly, healing would not happen between the tongue flap and the vomer bone, which also decreased the survival chance of the tongue flap after the operation. Thirdly, in this patient, the vomer was positioned in the middle of the palatal recipient site which was at the same level as the palatal shelves. It is important to note that the vomer was wide enough to keep the mucoperiosteal membrane on it to suture up the mesial side of tongue flaps bilaterally (Fig. 4). Although several modified tongue flap procedures had been reported in several previous studies, ^{5,27,28} to our knowledge, this is the first report proposing the use of double tongue flaps to repair hard palate fistula to date.

In the present case, we left the wound open on tongue dorsal surface instead of suturing up after lifting the tongue flap to prevent the tongue from shape deformation. Most of tongue flaps reported in previous studies were grafted unilaterally on one side of the median fibrous septum followed by a suture-up of donor sites. In our case, the wounds on dorsal surface of tongue were bilateral, making it impossible to suture donor sites up without causing deformity of the tongue or introducing major functional deficits.

There were several different surgical methods to managing palatal fistula such as: palatal flap, buccal mucosal flap, palatoplasty, facial artery myomucosal flap, and free tissue transfer. Nonetheless, it was widely accepted that tongue flap was the optimal solution indicated for the closure of large and scarred recurred palatal fistula.9 In this paper, we described a novel modified tongue flips procedure which was designed on the basis of the complex and unique condition of the patient. Specifically, a double tongue dorsal flap was applied to repair big anterior fistula as a complication after bilateral cleft lip and palate repair; developed septum sited at the same level as the palatal shelves on both sides which divided a huge fistula into 2 fistulas (Fig. 4); the mucousperiosteum on the inferior surface of septum was wide enough to be used.

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