

Up-to Date Review And Case Report

Oronasal fistula as a cleft palate sequela : closure with tongue flap. Case report with review of the literature

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Abstract – Introduction: Oronasal fistula have several etiologies. Most of the time, the origin is congenital associated with cleft lip and palate patients, or it can be acquired after trauma, infection, neoplasm, postsurgical complications or radio and chemonecrosis. Excepted for palatal obturators, their treatment is surgical and may use different intraoral flaps. **Observation:** A 9-year-old girl was sent to consult presenting an oronasal fistula as a sequela of a cleft lip and palate, initially treated in Kosovo (Albania). This was an anterior midpalate fistula, large around 2 cm, functionnaly responsible of a nasal speech and food reflux in the nasal airways. The closure has been done with a tongue flap. **Commentary:** Most of the oronasal fistulae secondary to a cleft palate are managed with palatal rotational flaps. Nevertheless, they cannot be used in some cases, especially when the fistula size is too large, the localization too anterior and with already scarred palatal tissues around. Then, tongue flap may represent a reliable alternative. **Conclusion:** The tongue flap allowed a complete closure of the oronasal fistula, with a very good tolerance from the child. It shows its interest into the list of the numerous existing oral flaps. However, considering the discomfort of this technique, it must be done in selected cases, with compliant patients.

Introduction

Oronasal fistulae are not a common reason for consultation. When the cause has been discovered, it is often a result of trauma, infection, tumors, postoperative iatrogenic side effects, or osteonecrosis [1]. The main etiology of oronasal fistulae remains congenital, associated with cleft palate [1].

Oronasal fistulae are congenital malformations caused by the anomalies of fusion of the different prominences involved in the embryogenesis of the face, between the fourth and tenth weeks of development *in utero*. They represent the most common congenital craniofacial malformation occurring in one in every 700 births [2].

Their management is performed by a multidisciplinary team and extends from birth to adolescence with a succession of surgical procedures. Nevertheless, the oronasal fistula frequently persists and is found in 15–35% patients after primary closure of the palatovelar slit [3,4]. Such fistulae may

cause speech disorders, regurgitation of food in the nasal cavity, bad odors, bad tastes, as well as upper respiratory tract infections [1].

The majority of oronasal fistulae are small and can be easily managed by palatal flaps.

Large fistulae are more complex because there is less healthy tissue available at the palatal level. Moreover, in cases requiring multiple surgical procedures, the scarring aspect of the palatal mucosa compromises the efficacy of any flaps used. Other flaps, such as a buccal mucosal flap, a buccal flap, a muscular-mucosal flap on the facial artery, tongue flaps, or even the free micro-braided flaps may be used [1,3]. Their respective indications are discussed in terms of the fistula width and location, and the patient's surgical history, the quality of tissues available, compliance, and dentition [1].

Observation

A 9-year-old patient came in for a maxillofacial surgical consultation for advice on the management of an oronasal fistula. The patient presented with a congenital bilateral cleft lip, which was treated through three procedures in Kosovo. The cleft lip was closed but alveolar continuity was found on the left side only.

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Fig. 1. Oronasal fistula as a cleft palate sequela: initial state.

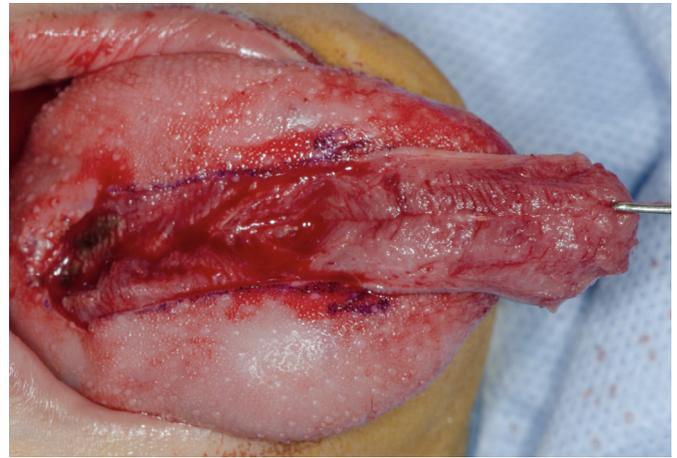


Fig. 3. Tongue flap lifting.

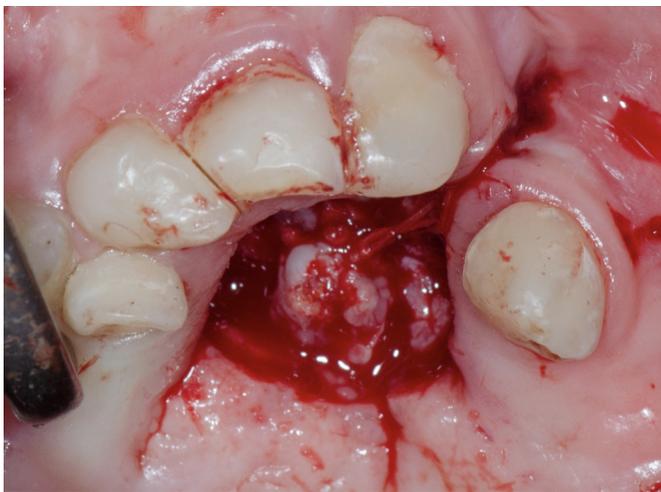


Fig. 2. First nasal layer.

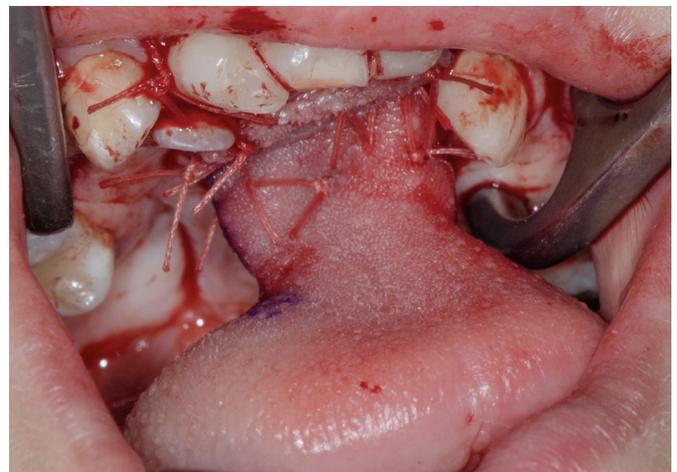


Fig. 4. Stitched tongue flap to the anterior part of the fistula.

At the palatal level, the patient had an extended oral communication of the primary palate to the velum, responsible for hyponasal speech and nasal reflux of liquids and solids. The first surgical intervention was to close the palate using rotational flaps. Subsequently, an oronasal fistula recurred between the palate and the median retro-incisor, and it was 2 cm in diameter (type V on the Pittsburgh classification) (Fig. 1).

A tongue flap was made to fuse this oral fistula to the cleft palate. The procedure was performed under general anesthesia with nasotracheal intubation. The patient was placed in the dorsal decubitus position with the neck hyperextended.

Infiltration of the palatal and lingual mucosa was performed with 8 ml of xylocaine and adrenaline (1/100 000). Corticosteroids (methylprednisolone 1 mg/kg) and antibiotics (amoxicillin 2 g/day) were administered before surgery.

A hinged flap was placed at the level of the palatal fistula allowing the rehabilitation of the first nasal mucosal plane (Fig. 2). The lingual flap was attached to the anterior medial pedicle and adapted to the width of the fistula. It was lifted with a cold blade with a 5 mm thickness, which was long

enough to allow the suture of the flap to the palate without any tension (Fig. 3). Lingual hemostasis was carried out using bipolar electrocautery. The tongue was sutured at separate points with Vicryl 3.0. The flap was sutured on the anterior and lateral parts of the fistula, carrying out a second closure, on separate points with Vicryl 3.0 (Fig. 4).

The surgical procedure was simple, with negligible pain and without any complications.

Postoperative pain was managed using a combination of paracetamol and nalbuphine while the patient was hospitalized. Oral prednisolone at a dose of 1 mg/kg was administered for the first 2 days after surgery, and antibiotic therapy with amoxicillin (2 g/day) was prescribed for 7 days. The food was administered orally, and it was a mixture of liquid and mixed foods, and the patient was asked to communicate in writing. Local care was performed after each meal by the family, by a simple rinsing with saline solution. The patient was discharged 48 h after the procedure, analgesia with level-1 analgesics was tapered off and the patient could eat. The patient has coped with this period between the two operating times very well.



Fig. 5. Flap healing one month after surgery.

The pedicle section of the flap was completed in the second phase of treatment. It was performed under general anesthesia after a period of 2 weeks following the first surgery.

An incision was made at the base of the lingual flap, thus releasing its anterior part.

Lingual hemostasis was performed using bipolar electrocautery. The flap was elongated. Hermetic sutures on the posterior part of the palatal fistula were made by separate points with Vicryl 3.0. Outpatient treatment required no additional procedures. The postoperative follow-up evaluations were very satisfactory with good healing 1 month after surgery at the level of the palatal fistula (Fig. 5) as well as at the level of the lingual mucosa (Fig. 6). The last follow-up examination, performed 6 months after the procedure, ruled out fistula recurrence, and the patient no longer had nasal reflux when eating. An improvement of the phonation was possible thanks to speech therapy.

Comment

In 1901, Eiselberg was the first to describe a pedicle tongue flap for use in the reconstruction of intraoral volume defects. Since then, the tongue flap has been used in several procedures for the reconstruction of the loss of volume from the intraoral defects, including the repair of oronasal and sinus connections and defects in the base, oral mucosa, lips, tongue, and oropharynx [1,5].

Its main limitation is the patient's tolerance, which is a determining factor for the success of the technique. Thus, the primary contraindications for tongue flaps are psychiatric disorders, mental retardation, and patients with heavy comorbidities, which can affect the efficacy of general anesthesia [5]. Active smoking, unbalanced diabetes, or severe malnutrition, are secondary contraindications that need to be addressed.

The first step is always performed under general anesthesia, with nasotracheal intubation. The resection of the pedicle can be performed under local anesthesia but requires a good patient compliance to achieve hemostasis in good conditions. In a young child, as in the case presented, it is necessary to perform this second procedure under general anesthesia.



Fig. 6. Tongue healing one month after surgery.

The postoperative procedures are relatively simple and common to other lingual surgeries with the immediate risks during the postoperative period being pain, edema, bleeding, infection, hematoma, and temporary hypoesthesia [5]. The main complications are the premature detachment of the flap, necrosis, speech impairment, and donor-site complications. Complications are often associated with improper application of the technique (flap too short sutured on, rotation too large, poor flap design) [5,6]. The tongue flap does not cause speech impediments when tongue integrity is not compromised [7].

Technically, it is accepted that the tongue-flap thickness must be 20% wider than the fistula because of a certain degree of flap retraction [8] and its design may be adapted specifically to the fistula shape [9]. Most surgical teams prefer a two-plane closure. The reconstruction of the nasal plane, either mucosal from the periphery of the fistula, or from a vomerine flap [10], allows better support and better flap stability, thus decreasing the risk of failure.

Alsaman *et al.*, however, showed a success rate of 100% with the closure of a palatal fistula using a tongue flap in a single procedure on a series of five cases, where technically the total closure of the nasal plane was not feasible [11].

In 1972, Jackson suggested the first use of the tongue flap for the closure of the palatal fistula in children with cleft deformities and showed that the flap was safe and well tolerated by children [12]. Piggot *et al.* also demonstrated the success of the technique in both adults and children, including 100% success in their group for children aged <2.5 years [13].

A literature review reveals that the majority of oronasal fistulae in cases of cleft palates are successfully managed by local palatal flaps, but success is limited in more complex situations [1,3,14].

Studies of the tongue flaps have been conducted in a few special situations:

- wide fistulae (>1 cm) [3,6,10,15-17];
- anterior fistulae [6,10,15,18,19];
- residual fistulae on scars from multiple surgical interventions [5,15].

These three criteria are different in children and adults, studies on the closure of oral fistulae using tongue flaps all report a high success rate of 85–100% with reliability and reproducibility [6,10,13,15–17,20].

Studies that have studied the result on phonation report 75% success rates in decreasing hypernasal speech [6,13,19]. The closure of an oral fistula by tongue flap remains effective if a transverse palatal expansion is performed, as shown in the series by Guzel and Altintas, with a 90% absence of recurrence [10].

Conclusion

Oral fistulae are the most common sequelae of patients with cleft palate history. Often when the fistulae are of moderate size, they are treated successfully with palatal flaps. However, when the fistula is wide or in an anterior position and/or when the tissues are scarred, palatal flaps cannot be used. In the case presented and the study of the literature show very good success rates with the tongue flaps, including success rates in the closure of the fistula, the improvement of phonation, or the stability of the closure after expansion.

Tongue-flap surgery, because of its technique and patient discomfort between the two surgeries, must be used in ideal conditions and on carefully selected patients by a surgeon who has mastered the technique.

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