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ORIGINAL ARTICLE**Anterior-based Tongue Flap for repair of recurrent wide anterior palatal fistula, refreshment of the technique and the outcome.**

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Submit Date 2020-07-22**Revise Date** 2020-09-14**Accept Date** 2020-09-30**ABSTRACT**

Purpose: The most complicated palatal operation is trial of closing large recurrent palatal fistula with previous attempts of closure, because of deficient local tissue which is fibrotic and relatively immobile, repeated surgery may jeopardize blood supply of the palatal tissue. The present study aims at evaluating the outcomes of using an anterior-based tongue flap in closure of recurrent wide palatal fistula.

Methods: Fifteen children with wide anterior palatal fistula after previous failed trial for closure using local palatal tissues were included in the study. Patients were managed by anteriorly-based tongue flap to be separated after 2 weeks. Post-operative data were recorded regarding flap viability, recurrence, flap bulk, speech difficulty and tongue disability.

Results: The mean age of included patients was 7.5 years. No intraoperative complications were recorded. Taste sensation and swallowing reflex were normal in all patients. Flap dehiscence and fistula recurrence was observed in one patient (6.66%). There was a statistically significant improvement in hypernasality post-operatively ($P < 0.01$). Nasal regurgitation was completely corrected in 13 out of 15 patients (86.66%).

Conclusion: Anterior-based tongue flap is an effective alternative technique for closure of wide anterior palatal fistula complicating palatal surgery and is associated with good functional outcome and patient satisfaction.

Keywords: Tongue flap, palatal fistula, recurrent fistula.

1. INTRODUCTION

One of the expected outcomes of cleft palate repair is to achieve complete separation of the nasal and oral cavities in addition to restoring the anatomical insertion of the palatal muscles to obtain normal function regarding speech. Failure of achieving complete structural integrity of the palate is recognized as an oronasal (palatal) fistula with persistent communication between oral and nasal cavities. The term, palatal fistula, is normally used for residual non-repaired cleft palate or breakdown of repaired palate [1].

Incidence of palatal fistula in secondary palate varies from 8.9 to 34%. Breakdown of palatal repair is one of the major causes of fistula formation, which is in turn may be related to amount of tension in the repair line, necrosis of the flap, greater palatine artery injury or mechanical trauma [2].

The most complicated palatal operation is closing a large recurrent palatal fistula after a previous failed attempt of closure, because of deficient local tissue which is fibrotic and relatively immobile. Repeated surgery may jeopardize blood supply of the palatal tissue with poor outcome of the repair trial.

Taking this into consideration, various techniques including local flaps and free flap utilization had been introduced for fistula repair but they lacked satisfactory outcomes. Lingual flap technique has been introduced since the beginning of the century [3]. The present study aims at evaluating the outcomes of using an anterior-based tongue flap in closure of recurrent wide palatal fistula as an alternative option for managing such patients.

2. PATIENTS AND METHODS

After obtaining Institutional Board Review (IRB) approval, fifteen children admitted to Zagazig University Hospitals with wide anterior palatal fistula after previous failed trial for closure using local palatal tissues in the period from July 2018 to July 2019 were included in the study as a comprehensive sample.

Written informed consent was obtained from all participants' parents. The study was done according to The Code of Ethics of the World Medical Association (Declaration of Helsinki) for studies involving humans.

Criteria for patient enrollment included size of fistula (>1x1cm), previous attempts of surgical repair with local tissue flaps and post cleft palate repair fistulae. Patients with traumatic or iatrogenic fistulae were excluded. All patients were operated by the same surgeons, managed and treated equally regarding the post-operative care, frequency of follow-up and management of complications if present.

2.1 Preoperative

After being evaluated by speech therapist preoperatively to evaluate the degree of hypernasality and speech affection, oral antiseptic gargle was prescribed for all patients three days before the operation date. Prophylactic third generation cephalosporin's was administered with induction of anesthesia.

2.2 Operative Technique

Under general anesthesia with endotracheal tube passed nasally, which is preferable, or orally and fixed to not interfere with the operative field, patient is positioned in supine position, shoulders elevated and head fixed, the mouth is opened using Dingman mouth gag.

The edges of the fistula are infiltrated with 1:200,000 epinephrine. The oral mucoperiosteum is incised around the edges of the fistula creating a flap for closure of the nasal side of the fistula (turn-over flap) with undermining of the oral mucoperiosteum for about 5 to 10mm for easy suturing with tongue flap edges.

The size of the fistula to be covered is measured and labelled on sterile sheet that applied on the dorsal surface of the tongue.

After marking the flap, a U-shaped incision is made along the dorsal surface of the tongue with its base directed anteriorly. The posterior limit of the flap is the circumvallate papillae and the anterior limit is one to two centimeters from the tip of the tongue. The thickness of the flap includes mucosa and a superficial layer of the muscles of the tongue (5-7mm to avoid a bulky flap interfering with swallowing and articulation after separation) and the width of the flap was decided according the width of the fistula with a maximum width of two thirds of the tongue width.

The flap is dissected and the defect in the tongue is closed primarily by full thickness absorbable sutures.

The tongue flap is turned up and edges of the flap are sutured to the mucoperiosteal edges of the fistula with polygalactin 910 sutures (Vicryl 4/0) using parachute technique with the mucosal surface of the tongue flap looking downwards (Fig. 2).

2.3 Post-operative care

Postoperatively, liquid diet is started at night of surgery and continued for the next 2 to 3 weeks till flap separation is scheduled and oral anti-fungal is started and continued till time of separation.

Intermaxillary fixation was not done as the child is old enough to be aware about the danger of traumatic flap separation. The parents and child were strictly instructed to avoid aggressive mouth opening and scheduled after two to three weeks to take down the flap (Fig. 3).

2.4 Flap Separation

The 2nd stage was done after 2-3 weeks under general anesthesia with the endotracheal tube passed nasally. The flap was taken down

using cutting diathermy, no more stitches were needed to fix the flap to oral mucosa, only hemostatic stitches on the dorsum of the tongue.

Oral fluid is allowed from night of operation; soft diet was commenced at 4th to fifth day then regular diet is started after one week (Fig. 4).

2.5 Follow up

Patients were followed up in Pediatric Surgery outpatient clinic weekly for one month then monthly for three months. Patients were evaluated in each visit for signs of fistula recurrence, the bulk of the flap, speech difficulties and tongue disability.

All patients were examined by speech specialist one-month post-operative for evaluation of speech improvement and the degree of hypernasality.

2.6 Statistical Analysis

The collected data were computerized and statistically analyzed using SPSS program (statistical package for social science) version 25.0. Qualitative data were represented as frequencies and relative percentages, while quantitative data were expressed as mean \pm SD (Standard deviation) and range. Wilcoxon Signed-Rank test was used for comparison of categorical ordinal variables. P-value was considered significant if $p < 0.05$ and highly significant if $p < 0.01$.

3. RESULTS

During a twelve-months period, a total of fifteen patients with recurrent fistula after palatal repair were included in the present study. The age of patients ranged from 4 to 12

years. There was a slight male predominance (60%). Most patients had a fistula less than 2cm² in size (73.33%) while only 4 patients had a fistula larger than 2cm² (Table 1).

The mean operative time was 53.2 \pm 6.2 minutes and the mean hospital stay was 1.45 \pm 0.53 days (Table 2). No intraoperative complications or events were recorded and no blood transfusion was required for any patient. Taste sensation and swallowing reflex were preserved in all patients.

Postoperatively, nasal regurgitation was completely corrected in 13 out of 15 patients (86.66%) while 2 patients only had persistent, yet decreased, nasal regurgitation (Table 3).

Fistula closure was successful in 93.33%, while on the other hand, flap dehiscence occurred at one side in 1 patient (6.66%) and separation was delayed till after 8 weeks to ensure adequate viability before interrupting the main blood supply from the tongue. After separation a smaller fistula persisted at the left side and scheduled for closure after complete healing (6 months) using the separated tongue flap.

Speech assessment was done pre-operatively and 1 month post-operatively by the same speech specialist for all patients. Evaluation items included nasal emission, hypernasality, and speech intelligibility. There was a highly significant statistical improvement in the degree of hypernasality post-operatively ($p < 0.01$) (Table 3). Overall, we had an excellent patient satisfaction in 86.6% of our patients.

Table (1): Demographic data and patients' characteristics.

Variable	Studied Patients (n=15)
Sex	
Male	9 (60%)
Female	6 (40%)
Age (years)	
Mean \pm SD	7.5 \pm 1.24
Range	(4-12)
Age Categories	
4-6	3 (20%)
6-8	7 (46.66%)
8-10	3 (20%)
10-12	2 (13.33%)
Size of Fistula	
1 to 2cm ²	11 (73.33%)
>2cm ²	4 (26.66%)

Table (2): Operative Course.

Variable	Studied Patients (n=15)
Operative Time (minutes)	53.2 \pm 6.2
Blood Loss (ml)	70.4 \pm 21.7
Hospital Stay (days)	1.45 \pm 0.53
Data are expressed as mean \pm SD	

Table (3): Follow-up Results.

Variable	Studied Patients (n=15)		
Follow-up (months)			
Mean±SD	4.46±0.74		
Median (Range)	4 (4-6)		
Nasal Regurgitation			
Corrected	13	(86.66%)	
Decreased	2	(13.33%)	
Fistula Closure			
Successful	14	(93.33%)	
Failed	1	(6.66%)	
Patient Satisfaction			
Excellent	13	(86.66%)	
Good	2	(13.33%)	
Fair	0	(0%)	
Poor	0	(0%)	
Hypernasality			
	Preoperative	Post-Operative	P-Value
Normal resonance	0 (0%)	2 (13.33%)	<0.01**
Mild hypernasality	3 (20%)	7 (46.66%)	
Moderate hypernasality	7 (46.66%)	5 (33.33%)	
Severe hypernasality	5 (33.33%)	1 (6.66%)	

** Highly significant using Wilcoxon Signed-Rank test

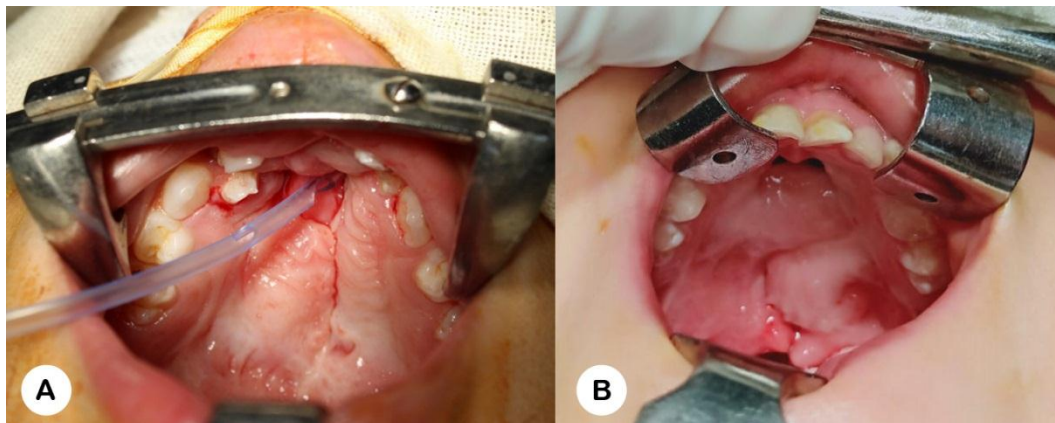


Fig. (1) Preoperative appearance of the palatal fistula in two patients. (A) A catheter is passed through the fistula into the nasal cavity. (B) Wide anterior fistula could be seen

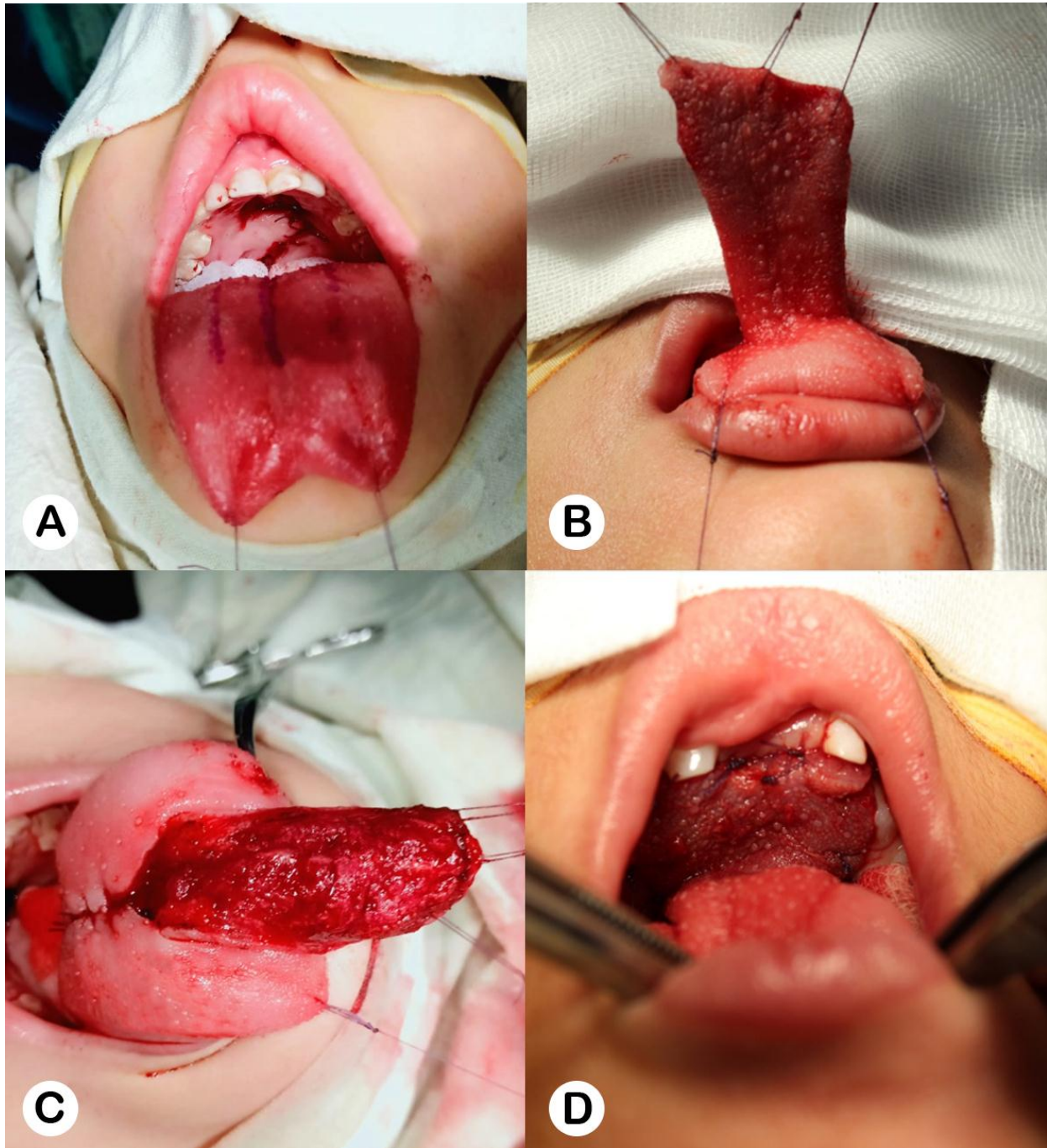


Fig. (2) Basic Steps of the technique. (A) Stay sutures and marking of the incision. (B) Dissection and elevation of the anteriorly-based tongue flap. (C) Hemostasis and primary closure of the tongue. (D) Final Appearance after suturing the tongue flap to the palate



Fig. (3) Two-weeks post-operative appearance of the flap with good viability

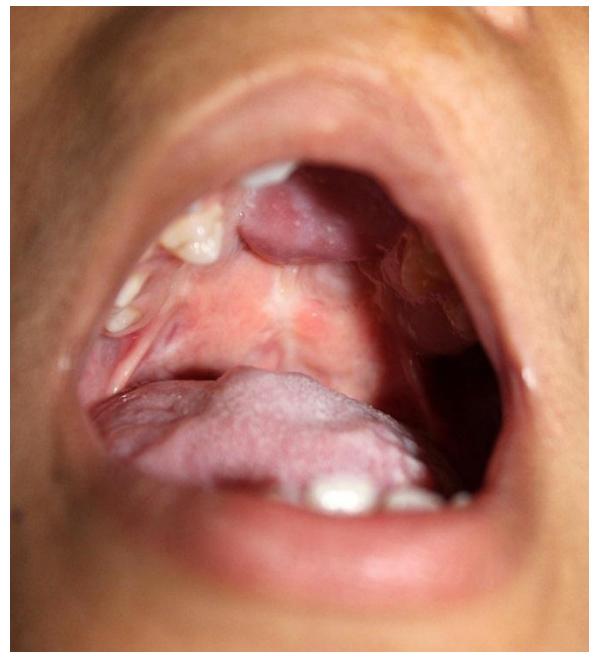


Fig. (4) One Month after separation of the flap

4. DISCUSSION

Palatal fistula is defined persistent communication between oral and nasal cavity regardless the size and shape of the defect. Fistula after palatal repair is predisposed by multiple risk factors including wide palatal cleft, repair under tension, vascular pedicle injury, hematoma or infection.

The guidelines of management of the palatal fistula is dependent on the type of cleft, site of

fistula, condition of surrounding tissue [4]. Minute fistula may be present without significant functional or cosmetic impairment which do not require management. Small sized fistula can be repaired using local palatine tissue with different closure artistry methods with high successful outcome. The true challenge facing the surgeons is closure of a giant and recurrent palatal fistula due to shortage of tissue and successful repair requires

transferring neighboring tissue for closure with ease and expected good results [4]. In the present study, an anterior based tongue flap has been used as an alternative to the deficient local tissue in cases of wide recurrent palatal fistula. Tongue flap has many advantages including being essentially tension free and don't require inter-maxillary fixation as the movement of the tongue is restricted by the resulting pain on excessive movement as well as the fact that most patients would have a tongue flap procedure at an age old enough to cooperate and understand the risk of traumatic separation of the flap. Another advantage is the rich blood supply supplying the tongue from the lingual artery and the extensive anastomosis provide good viability of the flap tissue [5].

The first reported use of a lingual flap to repair a fistula in the palate was presented by Gerrero-Santos and Altamirano in 1966. Since then, multiple authors reported successful trials of the tongue as a pedicled flap with abundant tissue to use for repair with a reported success rate ranging from 85% to 95.5% [4,7-10]. The reported parameters for successful flap repair included sufficient length of the flap (5 to 6 cm), a flap width somewhat larger than the defect, and a flap thickness of about 0.5 cm [11]. The present study showed a comparable success rate with 14 out of 15 patients repaired successfully (93.33%). Only one case suffered from partial dehiscence of the flap with small fistula recurrence.

The reported complications in literature are few. Busić et al. [11] reported partial marginal necrosis after division of the pedicle in one patient, another had complete necrosis after division of the pedicle, and another had complete necrosis of the distal part of the flap. Mahajan et al. [12] in a series of 41 patients reported bleeding in only one patient (2.43%) on the 9th post-operative day which was managed by separating the flap and hemostasis. Separation of the flap was scheduled within 10-21 days by most authors [4,13,14]. A worth mentioning challenge during the anesthesia for flap separation is the insertion of the

endotracheal tube as the tongue obscured the view of the laryngeal inlet and interfered with the insertion of the laryngoscope. This was overcome by inserting the tube nasally or gentle lateral traction of the flap by the laryngoscope during oral intubation. In our series, no adverse events were encountered by the anesthesiologists during intubation.

In all our cases, postoperative aesthetics (shape and symmetry) and functions (movement, taste and pain sensation) of the donor tongue site were preserved.

5. CONCLUSION

Anteriorly-based tongue flap is our currently preferred treatment option for management of recurrent palatal fistula complicating palate surgery. It is considered safe, highly effective with persistently good results with no mentionable complications and excellent patient satisfaction.

6. COMPLIANCE WITH ETHICAL STANDARDS

Funding: No funding was received for this study.

Conflict of Interest: All authors declare that they have no conflicts of interest.

Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent: Informed consent was obtained from all individual participants included in the study.

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