

Original Article

A retrospective analysis of incidence and management of palatal fistula

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ABSTRACT

Background: Cleft palate repair may be compromised by a number of complications, most commonly the development of a fistula. Fistulas may cause hypernasal speech, articulation problems and food or liquid regurgitation from the nose. **Objective:** The study determines the incidence and management of cleft palatal fistulas in a series of primary cleft palate repair surgeries. It is a retrospective analysis of total 185 palatal fistula cases operated at our hospital from the year 2004 to 2016. **Subjects and Methods:** Of 185 palatal fistulas, 132 cases had been operated at our institute for primary palatoplasty, and the rest 53 were the outside-operated cases. The patients with bilateral as well as unilateral cleft lip and palate were included. Isolated cleft palate patients were also included in the study. Palatal fistulas were subdivided into three types depending on their size. Anterior palatal fistulas were mostly treated by using tongue flap (65.57%), followed by local flaps (34.43%). Middle and posterior palatal fistulas were mostly treated by von Langenbeck Palatoplasty. One patient (>5 mm fistula) was treated using free radial forearm flap. **Results:** Anterior palatal fistulas (65.57%) were most commonly reported, followed by middle (24.86%) and posterior (9.18%). Most commonly, the size of the fistulas ranged from 2 mm to 5 mm. The complication rate was reported to be 3.75% in case of tongue flap and 11.9% complications were reported in case of local flaps. **Conclusion:** Tongue flap remains the flap of choice for managing very difficult and challenging anterior palatal fistulas compared to local flaps.

KEY WORDS

Cleft palate; palatal fistula; palatoplasty; tongue flap

INTRODUCTION

This study was conducted to evaluate the incidence and management of palatal fistulas in patients with cleft palate. Cleft lip and palate is one of the

most common congenital anomalies worldwide affecting babies 1:700 live birth.^[1] Cleft lip and palate together occur in every 1:1289 live births, cleft lip alone occurs 1:1000 live births, more commonly among the boys and

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three times more frequent than the cleft palate alone. Cleft palate alone occurs 1: 2500 live births with a higher incidence among the female babies.^[2] Cleft lip and palate is the most frequent diagnosis accounting for 46% of all the cleft populations.

Cleft palate repair aims to attain the development of normal speech without significantly impairing maxillary outgrowth, as well as minimising hearing loss and middle ear complications.^[3] In managing patients with cleft palate, the most controversial issues include the timing of surgery, speech development and facial growth.^[4] The ideal age for cleft palate surgery is usually 9–18 months. Speech and hearing are improved by cleft palate repair before 24 months of age. Delayed closure (after 5 years) is associated with retarded growth of the maxillofacial region.

The incidence of fistulas after palatoplasty ranges from 3% to 38%.^[5] Larger studies report rates in the range of 10%–20%.^[6] Palatal fistulas may present as asymptomatic holes or may cause such symptoms as speech problems, nasal regurgitation of fluids or difficulty in maintaining oral hygiene. The most common locations for fistulas are at the region around the incisive foramen, at the posterior nasal spine and the uvula.

All post-operative fistulas are found to be contributed to either failure of healing or breakdown of the original cleft palate repair. The incidence is highly variable although the primary cause remains the same in most, which is due to closure under tension and infection.^[7] Failure of healing of the palatal wound post-repair may lead to scarring and fistula.^[8] Anatomically, the cleft size as well as the technique of repair are factors which influence fistula occurrence.^[9] Cleft size affects the difficulty of surgical repair and thus, indirectly affects post-operative maxillary growth.^[10] Facial and palatal growth retardation following cleft repair is said to be due to the destruction of blood supply and scar formation.

SUBJECTS AND METHODS

The study determines the incidence and management protocol of cleft palatal fistulas in a series of primary cleft palate repair surgeries. It is a retrospective analysis of total 185 palatal fistulas out of which 132 cases had been operated at our institute for primary palatoplasty, and the rest 53 were the outside-operated cases [Table 1]. The

technique for primary palatoplasty at our institute was von Langenbeck palatoplasty and two-flap palatoplasty with radical muscle dissection and posterior sling formation. The incidence of palatal fistulas and their management by various methods has been discussed. The gender and the age predilection has also been discussed, laying stress on the location and size of palatal fistulas along with the rate of complication.

The clinical records of the patients with palatal fistulas who underwent fistula repair between 2004 and 2016 were retrospectively reviewed. This included 132 cases operated at our institute for primary palatoplasty, and the rest 53 were the outside-operated cases. Patients included are those who had a complete operative and follow-up medical records with a minimum follow-up period of at least 2 months. Other variables of interest are noted for each patient, which include gender, age, type of cleft, location and size of fistula, method of repair and complication if any reported. The patients with bilateral as well as unilateral cleft lip and palate were included. Isolated cleft palate patients were also included in the study. Fistulas could be single or multiple in number. Anterior palatal fistulas were considered to be in the anterior part of the hard palate, middle palatal fistulas at the junction of hard and soft palate, and posterior palatal fistulas were confined to the soft palate. Palatal fistulas were subdivided into three types depending on their size—small (<2 mm), medium (2–5 mm) and large (>5 mm).

RESULTS

Between 2004 and 2016, a total of 2060 patients were operated for primary cleft palate repair at our institute, out of which total 153 patients were diagnosed for palatal fistula. Of these 153 patients, 21 patients were not operated as 10 of them had pinpoint fistulas, which resolved with time and 11 of them did not turn up for follow-up and rest of 132 patients were operated for palatal fistula repair. Hence, the true incidence rate of palatal fistula in our series was 7.427% (153 out of 2060 patients). Out of a total number of 185 palatal fistulas operated at our institute, 122 (65.94%) were anterior palatal fistulas, 45 (24.32%) were middle palatal fistulas, 17 (9.18%) were posterior palatal fistulas and one very large fistula almost whole of the palate (0.54%). Outside operated cases had more number of anterior palatal fistulas, and

the size of the fistulas was similar to our study (mostly ranging from 2 mm to 5 mm).

In terms of age, the most common age group at which palatal fistula cases turned up for fistula closure was 6–10 years (23.78%), followed by <5 years (17.29%), 21–25 years (17.29%), 26–30 years (14.05%), 16–20 years (12.43%), 11–15 years (9.73%), 31–35 years (3.78%) and 36–40 years (1.62%).

The reason for less number of patients operated under the age of 5 years is because this series includes a large number of patients who reported late for palatal repair or palatal fistula repair (when palate has been operated outside). Most of our own cases were taken up for palatal fistula repair after 6 months of palatoplasty.

In terms of gender, male predominance was reported as out of total 185 cases of palatal fistulas operated, 120 were male (64.86%) and 65 were female (35.13%).

Incidence wise, cleft palatal fistulas were most commonly seen in patients having bilateral cleft lip and palate (21.14%), followed by unilateral cleft lip and palate (4.17%) and isolated cleft palate (3.79%) [Table 1].

Fistula rate was found to be maximum in case of bilateral cleft lip and palate patients (21.14%) whereas it was comparatively less in case of unilateral cleft lip and palate patients (4.17%) and incomplete cleft palate patients (3.79%) as shown in Table 1. Number of cases operated for anterior palatal fistula were 122, middle palate fistula was 45, posterior palatal fistula was 17 and one case of large palatal fistula involving almost whole of palate operated by radial artery forearm flap (RAFF) as shown in Table 2.

In terms of management, all palatal fistulas were segregated into groups based on location and size. Location wise, it has been divided into anterior, middle and posterior palatal fistulas, while according to size, it has been classified into <2 mm (small), 2–5 mm (medium) and >5 mm (large).

Out of 122 anterior palatal fistulas, 26 cases (21.31%) were of the size of <2 mm, 76 cases (62.29%) were of the size of 2–5 mm and 20 cases (16.39%) was of the size >5 mm.

Out of 45 middle palatal fistulas, 19 cases were of the size (<2 mm), 25 cases (56.53%) of the size of 2–5 mm and 1 case (2.17%) was of the size of >5 mm.

Table 1: Rate of fistula formation

<i>Diagnosis</i>	<i>Cases operated at our institute</i>	<i>Fistula formation in our cases</i>	<i>Percentage rate of fistula formation in our cases (%)</i>
Unilateral CLP	1342	56	04.17
Bilateral CLP	402	85	21.14
Incomplete CLP	316	12	03.79
Total	2060	153	

CLP: Cleft lip and palate

Table 2: Plan of fistula repair

<i>Palatal fistula</i>	
Operated at our institute primarily	132
Primarily outside operated	53
Total	185
Anterior palatal fistula	
Tongue flap	80
Local flap	42
Total	122
Middle palatal fistula	
Langenbeck palatoplasty	45
Posterior palatal fistula	
Langenbeck palatoplasty	17
Large palatal fistula almost involving whole palate	
Radial artery forearm flap	1
Total	185

Out of 17 cases of posterior palatal fistulas, 7 cases (41.18%) were of the size <2 mm, 9 cases (52.94%) were of the size of 2–5 mm and 1 case (5.88%) was of the size of >5 mm.

Most of the anterior palatal fistulas (80 cases) were closed using tongue flap [Table 2 and Figures 1-3] followed by local flaps (21 cases) which included local mucoperiosteal flaps or extended alveolar mucoperiosteal flap and mucosal flaps [Figures 4 and 5]. Twenty cases of anterior palatal fistulas were closed using redopalatoplasty by two flap technique [Figures 6-8]. One case of anterior palatal fistula of size 2–5 mm was operated using buccal mucosal flap [Figures 9-11].

Of 80 cases of anterior palatal fistulas closed by tongue flap [Figures 1-3], we encountered dehiscence in 3 patients (3.75%). All the three patients were of the age <5 years and were further managed by flap reattachment. On follow-up, these cases healed well. In this way, tongue flap was considered as a successful treatment of choice for managing challenging anterior palatal fistulas hard to repair.^[11]

Middle and posterior palatal fistulas were treated by redo-palatoplasty using von Langenbeck technique [Figures 12 and 13].

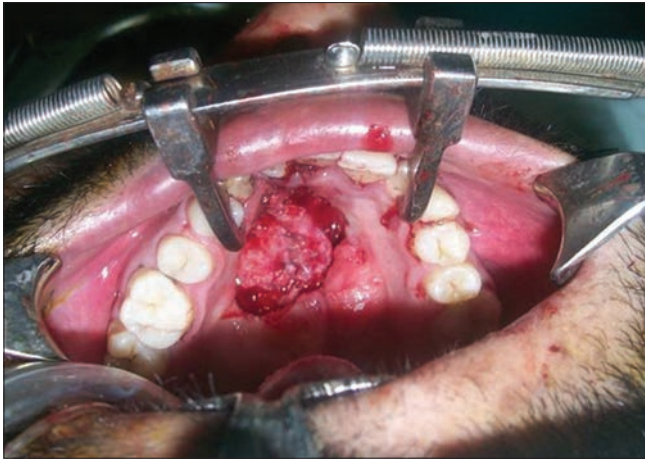


Figure 1: Anterior palatal fistula >5 mm



Figure 2: Tongue flap attachment

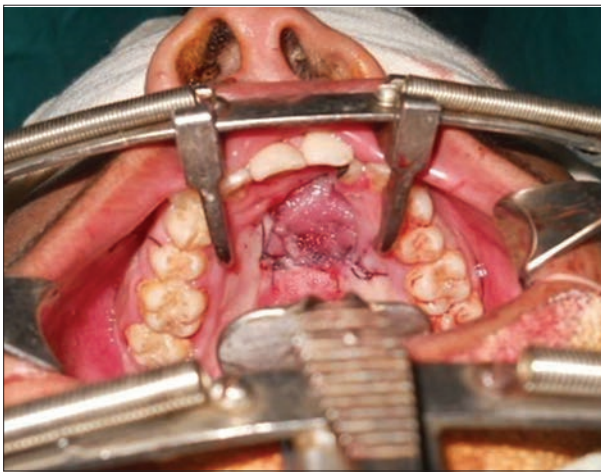


Figure 3: Tongue flap for anterior palatal fistula closure after division and inseting

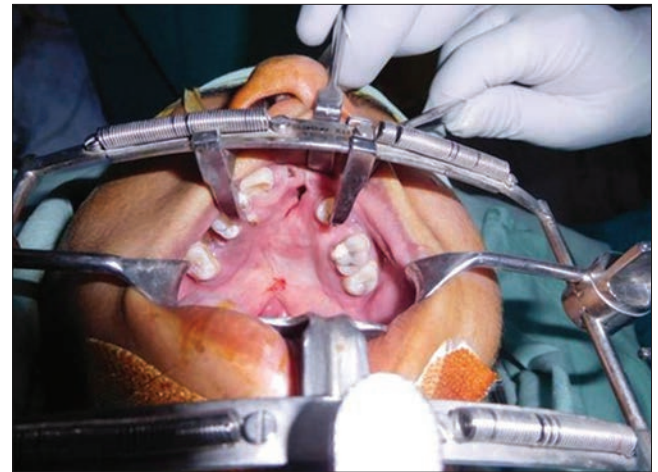


Figure 4: Anterior palatal fistula 2 mm to be corrected by using local flap (extended alveolar mucoperiosteal flap)



Figure 5: Fistula closure by local flap - extended alveolar mucoperiosteal flap

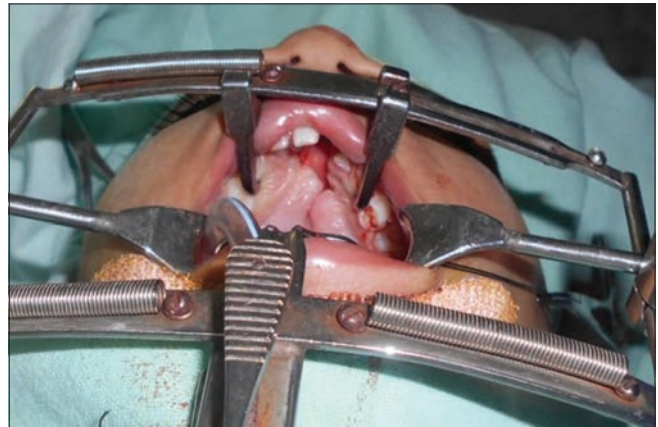


Figure 6: Anterior palatal fistula to be corrected by two flap palatoplasty

One case of very large palatal fistula involving almost the whole of the palate was treated by RAFF [Figures 14-16].

The complication rate for tongue flap was reported to be 3.75% (3 cases), due to flap dehiscence in three patients, which required reattachment. The recurrence rate was noted in these patients after 6 months of follow-up. In case of local flaps, the complication rate was 11.9% (5 cases)

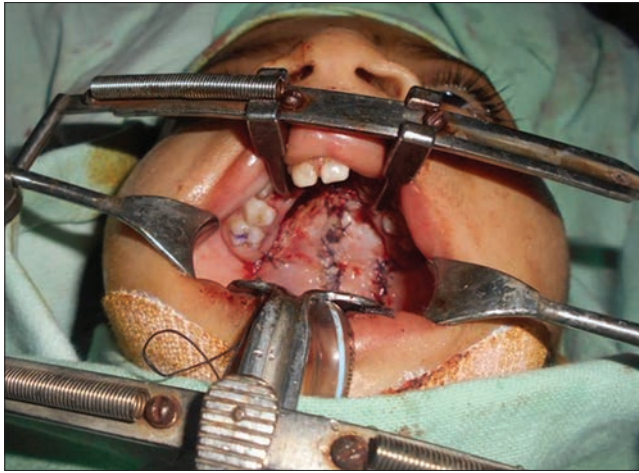


Figure 7: Two flap palatoplasty for fistula closure



Figure 8: Postoperative follow-up (2 months)

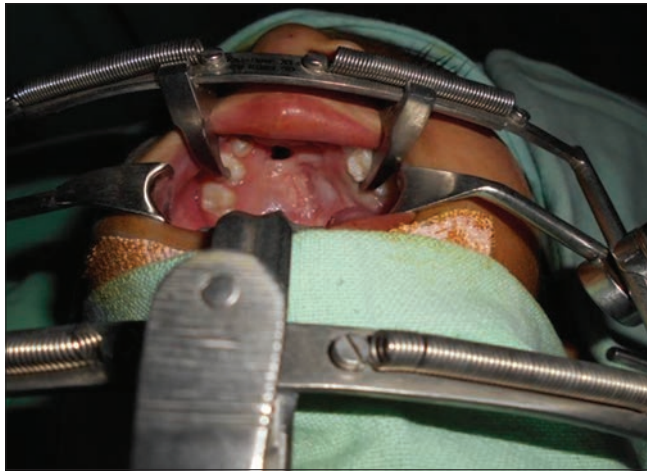


Figure 9: Defect of anterior palatal fistula to be corrected by buccal mucosal flap

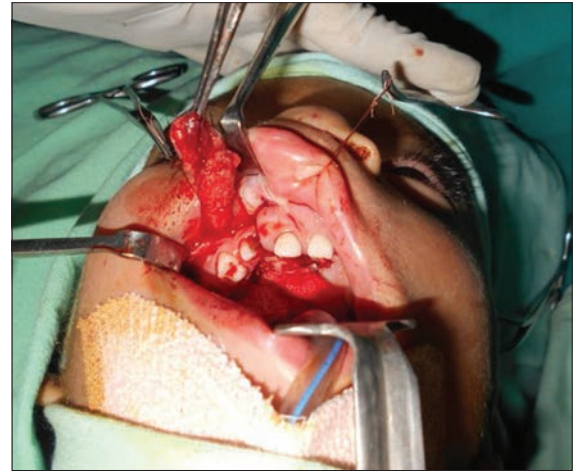


Figure 10: Harvesting of buccal mucosal flap

out of which two cases were of two flap palatoplasty. These were further managed by redo-palatoplasty and tongue flap in case of comparatively large palatal fistulas. In case of middle palatal fistulas complication rate was 8.69% (4 patients), and in case of posterior palatal fistulas, it was 5.88% (1 patient).

DISCUSSION

Despite improved technique of repair of cleft palate, fistula occurrence is still a possibility either due to an error in surgical technique or due to poor tissue quality of the patient. The incidence of palatal fistulas is more common in bilateral cases than unilateral ones, as reported by Musgrave and Bremner.^[8] Our study also reports a higher incidence of fistula in patients having bilateral cleft lip and palate (21.14%) as compared to unilateral cleft lip and palate (4.17%) and isolated cleft palate (3.79%).

About 3.75% of cases operated by tongue flap had residual fistulas due to flap dehiscence. These were further managed by tongue reattachment. The post-operative period of follow-up was 6 months.

Michael H. Carstens^[11] conducted a study encountering anterior palatal fistula in 77.5% of cases. In most of the cases, this results from failure to achieve control of the anterior nasal floor. We also report a high incidence of anterior palatal fistulas (65.94%) as compared to middle palatal fistulas (24.32%) and posterior palatal fistulas (9.18%). One very large palatal fistula, almost involving the whole of the palate was also reported.

In case of anterior palatal fistulas, the most common technique used for closure included local flaps (extended alveolar mucoperiosteal flaps and buccal mucosal flaps), two flap palatoplasty and tongue flap. The condition of native tissue was a good factor which helped the



Figure 11: Result after inset of flap and closure of fistula palatoplasty

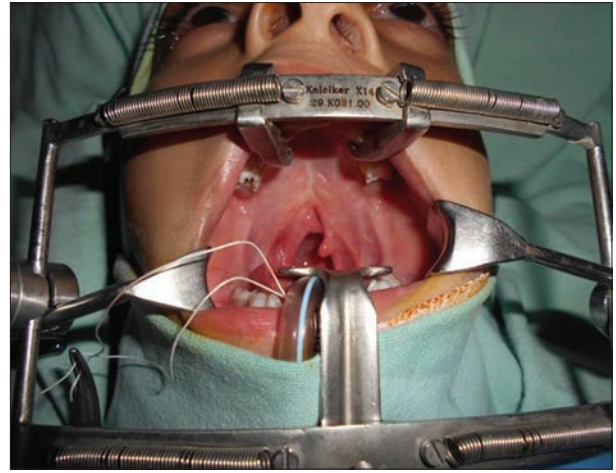


Figure 12: Soft palatal defect to be corrected by langenbeck palatoplasty

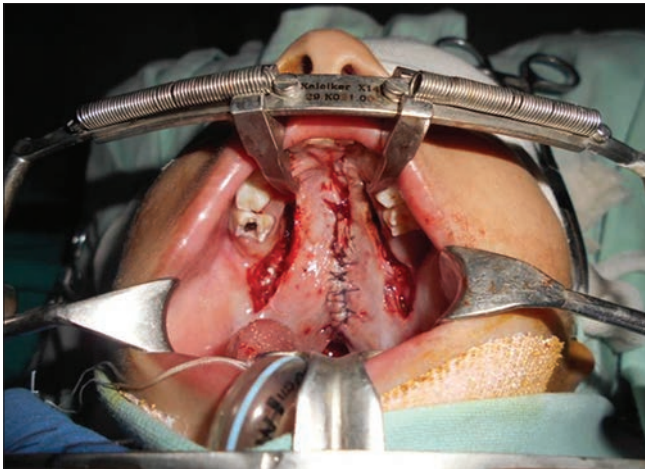


Figure 13: Langenbeck palatoplasty for correction of defect



Figure 14: Radial forearm free flap: Large palatal fistula involving almost whole of palate



Figure 15: Radial forearm free flap: Fistula closure by radial forearm flap



Figure 16: Radial forearm free flap: Post-operative result

surgeon to decide the technique to close similar types of fistulas. Local flaps were used only in those cases where the native tissue (with rugae) was available. Otherwise, we prefer bringing virgin tissue from other

areas. Local flaps were mostly done in cases <2 mm of size and where the condition of native tissue was favourable for the closure of fistula. In case of scarred tissue, compromised vascularity and fibrosis, the local

flap cannot be easily mobilised, and chances of flap necrosis are high.

Although commonly the anterior palatal fistula closure is established by the use of local flap at times, the size and site of fistula make use of local flap for its repair a remote possibility. Hence, for anterior palatal fistula >2 mm size, tongue flap was used as a workhorse flap in our series of patients. The use of tongue flap gave promising results in case of difficult anterior palatal fistulas with a shortage of tissue. The advantage includes the central position of tongue in the floor of mouth facilitating mobility and positioning of the flap. Success depends on proper flap elevation, tension-free nasal layer closure, edge-to-edge approximation of flap with palatal tissues and good tension-free primary closure of donor area near base of the flap.^[12] Middle and posterior palatal fistulas were mostly treated using von Langenbeck palatoplasty. Radial artery forearm-free flap was used in case of very large palatal fistula. Although Cohen *et al.* reported recurrence rate of 37%,^[13] our study shows a very low rate of 7.427% which is in concordance with the study of Musgrave and Bremner^[8] with a recurrence rate of 7%. Many authors have published very different data about the incidence of oronasal fistulas. The lowest incidence (0%) was published by Stewart *et al.*,^[14] followed by 0.7% by Brusati and Mannucci,^[15] 0.76% by Losee *et al.*,^[16] 3.4% by Wilhelm *et al.*,^[17] 3.6% by Khosla *et al.*,^[18] 4.7% by Inman *et al.*,^[19] 8.7% by Muzaffar *et al.*,^[20] 12.8% by Phua and Chalain,^[21] 15% Sommerlad.^[22] The overall recurrence rate of fistula was nil in case of tongue flap whereas it was 11.9% in case of local flaps, 8.69% in case of middle palatal fistulas and 5.88% in case of posterior palatal fistulas. In our series of tongue flap patients, only three cases of flap dehiscence were noted, which were managed further by reattachment of flap. None had a residual fistula in 6 months of follow-up. Hence, tongue flap was successfully used in managing challenging anterior palatal fistulas.^[12,23,24]

CONCLUSION

Tongue flap remains the flap of choice for managing very difficult and challenging anterior palatal fistulas, compared to local flaps. For middle and posterior palatal fistulas, von Langenbeck palatoplasty gave good results. For very large palatal fistulas, free flap (radial forearm flap) makes a good choice.

Declaration of patient consent

The authors certify that they have obtained all appropriate

patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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