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Case Report | Plastic Surgery

# A RARE CASE OF CONGENITAL PALATAL FISTULA WITH NORMAL SOFT PALATE AND UVULA


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| **Sushrut Tated*** | Senior Resident, Department of Plastic Surgery, Jawaharlal Nehru Medical College and Hospital, Aligarh, Uttar Pradesh, India*Corresponding Author |
| **Arshad Hafeez Khan** | Professor, Department of Plastic Surgery, Jawaharlal Nehru Medical College and Hospital, Aligarh, Uttar Pradesh, India |
| **Asif Iqbal Shaikh** | Senior Resident, Department of Plastic Surgery, Jawaharlal Nehru Medical College and Hospital, Aligarh, Uttar Pradesh, India, |


**ABSTRACT** Cleft lip and palate are commonly observed congenital anomaly. But congenital palatal fistula is very uncommon. Only few cases have been reported in literature globally. Irrespective of the doubts in etiology and pathogenesis, due to its rarity it is important to report these rare cases in the literature as and when it comes to the notice of the clinicians. Herein,we present a case of two-year-old child presented to us with isolated congenital palatal fistula. Apart from nasal regurgitation on feeding and URTI, all other things were normal. Midline, rectangular fistula of 2 cm x 1.5 cm extending from incisive foramen and involving posterior edge of hard palate was seen. Patient responded well to the surgery and postoperative period was uneventful.

**KEYWORDS :** Palatal fistula, soft palate, uvula

## Introduction

Cleft lip and palate are commonly observed congenital anomaly. But congenital palatal fistula is very uncommon. Usually it has been found to be associated with submucous cleft palate. Further isolated congenital palatal fistula without submucous cleft palate is even more rare.Congenital fistula was first reported by Veau V et al in1931. [1]

Till now only 30 cases of the congenital palatal fistula have been reported. [2] Due to this rarity, it is difficult to estimate the incidence of this condition. But even then, it has been estimated that the incidence could range between 0.17-0.45% of all the cleft palates and 6-17% among those who present with submucous cleft palate. [3]

It is also not possible to explain the palatal fistula formation on the basis of embryological theory of mesodermal migration. [4]

There is disagreement as to whether this congenital fistula of the hard palate is due to trauma or as a result of malformation. Some argue that it is due to the trauma during pregnancy or after birth to the submucous cleft palate and some argue that it is due to malformation of submucous cleft. [5]

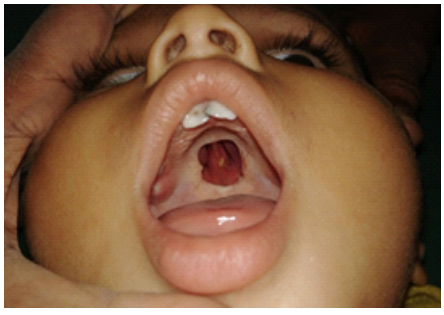
Irrespective of the doubts in etiology and pathogenesis, due to its rarity it is important to report these rare cases in the literature as and when it comes to the notice of the clinicians. Here we present a rare case of congenital palatal fistula not associated with submucous cleft palate in a two-year-old child.

## Case Report:

A two-year-old male child presented to our outpatient department with palatal fistula. Parents were unable to recall any history of trauma. Antenatal period was normal. It was an institutional normal vaginal delivery with normal birth weight of the child. There was no family history of cleft lip and cleft palate in any of the family members. At the time of presentation, the child was having upper respiratory tract infection. Pigeon bottle was used for feeding the child and it was observed that nasal regurgitation was present. During sleep, snoring was not observed.

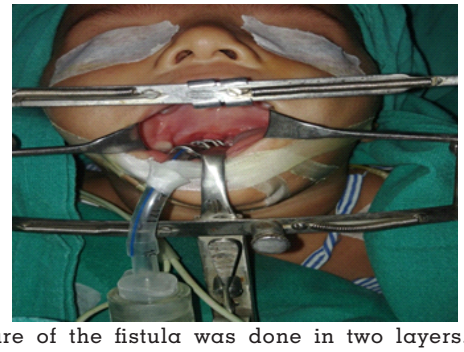
The fistula was midline. It was seen at the junction of hard palate and soft palate. The fistula was seen involving the posterior edge of the hard palate to incisive foramen. The fistula was about 2 cm long and 1.5 cm in width. Shape was rectangular. There was no any palpable depression in the soft palate. Vomer was underdeveloped and unattached to either palatal shelves. [Fig. 1]

**Figure 1: Preoperative photograph showing palatal fistula**



Parents consented for surgery and the child was taken for repair of the congenital palatal fistula under general anesthesia. Uvula was normal. [Fig. 2]

**Figure 2: Intraoperative photo showing normal uvula and soft palate**



Closure of the fistula was done in two layers. Oral mucoperiosteal flap were raised on both the sides from the undersurface of the palate. Nasal mucosal layers were developed on either side. However, the nasal layer was deficient in the anterior part.So, it was not possible to attain the full closure of the nasal mucosal layer in the anterior part. But it was possible to completely close the oral mucoperiosteal layer. [Fig. 3 and 4]

**Figure 3: intraoperative photo showing big palatal fistula and normal soft palate**

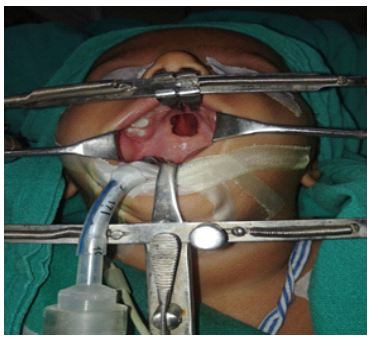
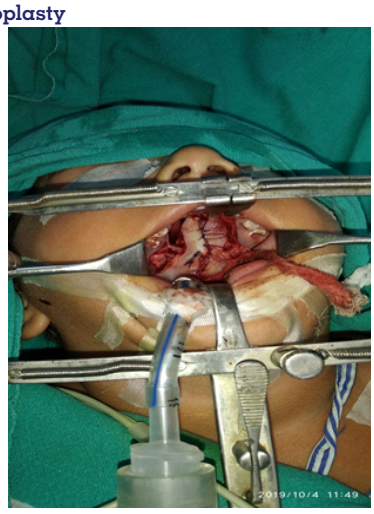
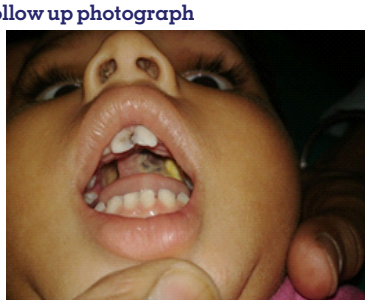


**Figure 4: Intraoperative photo after complete closure of two flap palatoplasty**




Postoperative day three, the patient was discharged and there were no complications. Follow up period was uneventful with no complaints. [Figure 5]

**Figure 5: Follow up photograph**



## DISCUSSION:

It is a common occurrence though not a rule that the congenital palatal fistula and submucous cleft palate are commonly associated. Lynch JB et al [6] in 1966 reported congenital palatal fistula not associated with submucous cleft palate. There was not history of trauma, occurred in children and the same location as reported in the present case report. Hence the cases reported and described by Lynch JB et al [6] are similar to our case.

Fara M [3] in 1971, reported four cases of palatal fistula and among them one was associated with submucous cleft palate. Cheng N et al [7] in 1998 reported four cases of congenital palatal fistula and submucous cleft palate was associated in all these cases. They stated that congenital palatal fistula is not due to any other factors but may be caused by failed differentiation in the palate during fetal life.

In 2003, Jagannathan M et al [8] presented a case of congenital palatal fistula with normally developed posterior palate, uvula with a normal-looking muscular sling. Author concludes that this condition could not be explained by any of the embryological theories and label this condition as an embryological enigma. This case was similar to our case.

In 2006, Rogers GFet al [2] reported a case of congenital palatal fistula with submucous cleft palate. Posterior part of soft palate was intact. Uvula showed thin raphe. The authors noted that muscles were hypoplastic.

Karacan M et al [9] in 2009 reported a case of isolated palatal fistula which was not associated with submucous cleft palate. Parents denied surgery. that their child will undergo surgery. They were lost to follow up for short duration. But at 18 months of follow up, they came with the child with spontaneous closure of the fistula. This was remarkable which made author to conclude that given time, such fistula may get closed spontaneously.

Eshete M et al [4] described two cases of congenital palatal fistula. In that one was associated with submucous cleft palate.

Most of cases published were associated with submucous cleft palate. [2] But our case was an isolated case of palatal fistula with normal soft palate and uvula.

## CONCLUSION:

Isolated case of palatal fistula with normal soft palate and uvula is rare. Appropriate management brings about the required successful outcome.

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