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RESEARCH ARTICLE

SUBMUCUS CLEFT PALATE WITH CONGENITAL ORO NASAL FISTLA: REPORT OF A RARE CASE

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ABSTRACT

Submucus Cleft Palate is a rare congenital disorder which is can present in paediatric age group in a variety of ways. Most of the submucus clefts are seen in posterior part of the palate. We report a very rare variety of submucus cleft with congenital oro nasal fistula at soft and hard palate junction. To the best of our knowledge no case has been reported.

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INTRODUCTION

Oronasal fistula is a known complication of cleft palate surgery. It's presence since birth is rare and very few cases of congenital oro nasal fistula in a case of submucus cleft palate have been reported. Ever since it's description by Roux in 1825 (Roux, 1930) Submucus Cleft Palate has been a challenge to manage. The triad of bifid uvula, Palatal Muscle diastasis and bony notch of the Hard palate in submucus cleft palate has been noted by Calnan (Porterfield, 1965). We present a case of 2 year female child who presented in Plastic Surgery outpatient department at Government Medical College, Nagpur with Submucus cleft palate with congenital oro nasal fistula.

MATERIAL AND METHODS

First born female child born of a non consanguineous marriage at full term of an uneventful vaginal delivery at hospital presented to us at 2 years of age with a defect in palate since birth. According to her parents, there was no history of similar disorder in her family members.

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There was no history of trauma. There was history of regurgitation of food particles in the nose during eating and drinking. The mother had no history of intake of any medication apart from supplementation of iron and folic acid. There was no history of exposure of radiation in the antenatal period. Age of mother at the time of her birth was 24 years. On examination, there was submucus cleft palate with a oro nasal fistula. Size of the fistula was 0.5 mm by 3 mm in maximum anteroposterior and horizontal dimensions respectively (Figure 1). She was investigated for anesthetic fitness and screened for any other congenital anomaly. No other congenital anomaly was present in the patient. She was examined from E.N.T. side and had normal hearing. She underwent pushback palatoplasty under general anesthesia. Post operative period was uneventful and she was discharged on day 8 in stable condition with healthy suture line. Nasal regurgitation was cured after the procedure. Speech therapy was started postoperatively. Patient was asked to follow up for review at regular intervals. Post operative follow up at 6 months showed no evidence of fistula healthy suture line.

DISCUSSION

Submucus cleft palate is a rare disorder with an incidence of 1:2500 to 1:6000 live births.

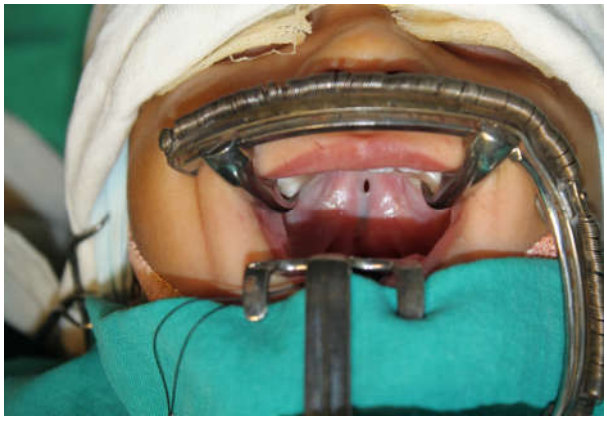

Figure 1. Preoperative photograph

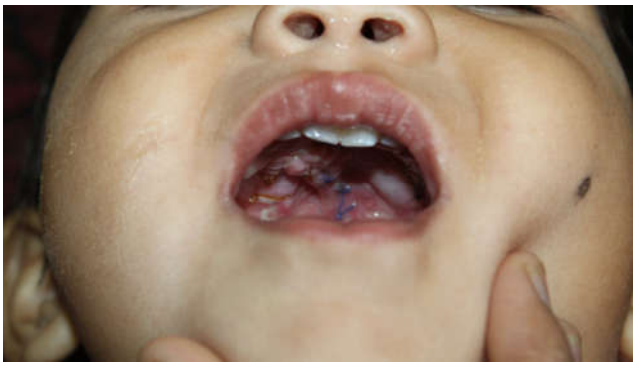

Figure 2. Postoperative photograph

Incidence of submucus cleft palate with oro nasal fistula is unknown (Gosain *et al*., 1997). It is one of the rarest type of facial clefts. Though most patients have normal speech, it has an effect on the development of feeding, speech, hearing and thus interfering with the social development of the child. Patients may take long feeding time. It can be associated with velopharengeal insufficiency which is associated with conductive hearing loss. Diagnosis is mainly clinical. Surgical treatment helps patients with problems with feeding and speech. Patient treated for velopharyngeal insufficiency have good outcomes (Husein *et al*., 2004). Several procedures have been advocated for the treatment of mucosal cleft palate ranging from Furlow's procedure and Pushback palatoplasty, minimal incision palatopharyngoplasty. A pharyngeal flap has been repeatedly observed to have appreciable correction in velopharyngeal insufficiency (Gosain *et al*., 1997). Post operative speech therapy should be started. Though Oro nasal fistulas have been reported in the post operative period after surgery for cleft palate, congenital oro nasal fistula in a patient of sub mucus cleft palate has been reported in very few patients. Rogers *et al* reported a case of congenital fenestration of palate in a newborn (Rogers *et al*., 2006). He has also mentioned that only five out of twenty six cases of submucus cleft palate had congenital oro nasal fistula. It is unclear whether this fenestration is due to trauma in pre or post natal period or is it a result of malformation in most cases (Rogers *et al*., 2006). Mekonen *et al* have recently reported a case of submucus cleft palate with congenital oronasal fenestration (Mekonen *et al*., 2016). Congenital fistula of the hard palate was first reported by Vaeu and Borel in 1931 (Mekonen *et al*., 2016). To the best of our knowledge, about 33 cases have been reported since then. The incidence of submucus palate in general population has been reported to be 0.02- 0.08% (Gosain *et al*., 1997). Perforation in submucus cleft palate can result in the prenatal period or after birth (Mehendale *et al*., 2003). It is very rare. It could be due to prenatal rupture of the submucus cleft palate (Mekonen *et al*., 2016). After birth, perforation in a submucus cleft palate can be the result of trauma or infection (Park *et al*., 2016) to the palate. Early postoperative period can be complicated with difficulty in respiration, bleeding and wound breakdown. Late postoperative complication may be in the form of fistula and velopharyngeal insufficiency (Chen *et al*., 1996). Patients of submucus cleft palate may have poor speech outcomes after Surgery (Park *et al*., 2016). Velopharyngeal insufficiency in patients of Submucus cleft palate in the preoperative period can be addressed in a better manner by Furlow palatoplasty as compared to pharyngeal flap surgery (Chen *et al*., 1996).

Conclusion

Submucus cleft palate with congenital oro nasal fistula is very rare. Diagnosis is clinical. Various treatment options are available of which pushback palatoplasty is a standard method in cases of minimal velopharyngeal insufficiency. A multidisciplinary approach should be used for better overall development of the child postoperatively for better outcome.

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