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CASE REPORT

A CASE REPORT OF PALATAL HAMARTOMA WITH BIFID TONGUE: RARE CONGENITAL ANOMALY

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ABSTRACT: Here is a rare case of congenital anomaly, palatal hamartoma with bifid tongue in a 5 month old infant with non-oral-facial-digital syndrome. Informant, mother complain of difficulty in sucking breast milk and dribbling of milk during breast feeding and swelling in the palatal part of the oral cavity, mucous cyst in the floor of the mouth. The palatal swelling excised and the bifid tongue was repaired in layers surgically. Diagnosis of palatal hamartoma was based on the histopathological report. The review of literature, the description of the lesion, the diagnosis and the management of this finding are outlined.

KEYWORDS: Hard Palate, Bifid tongue, Hamartoma.

INTRODUCTION: Hamartoma, a benign tumour like malformation composed of a disordered mixture of mature tissue that normally occur in the affected part, but with a predominance of one particular tissue.(1) Tongue develops during 4th week of gestation, in the floor of the primitive oral cavity. Two lateral lingual swellings and one median swelling Tuberculum impar from 1st pharyngeal arch forms the anterior 2/3rd of the body of the tongue.

The lateral lingual structures rapidly grow and cover the tuberculum impar to form the anterior 2/3rd of the tongue. When this process is disturbed, the tip of the tongue is divided longitudinally for a certain distance giving rise to cleft tongue or bifid tongue. They may occur as an isolated entity or a part of clinical syndrome with oral-facial-digital abnormalities.(2,3,4,5)

CASE REPORT: A 5 month old female infant presented with swelling in the mouth, slowly growing in size since birth. History of difficulty in swallowing and dribbling of the milk during breast feeding was present. There is no significant antenatal history (mother non-diabetic). Baby was delivered at term normally.

On examination baby was malnourished.

Tongue was bifid in its anterior 2/3rd completely. Mucous cyst present in the floor of the mouth at frenulum measuring about 3x3 cms. Big soft swelling over the hard palate measuring about 5x5 cms protruding through the mouth exteriorly. Extra oral examination revealed no congenital facial defects. Systemic examination reveals no congenital digital and other anomalies (Fig 1).

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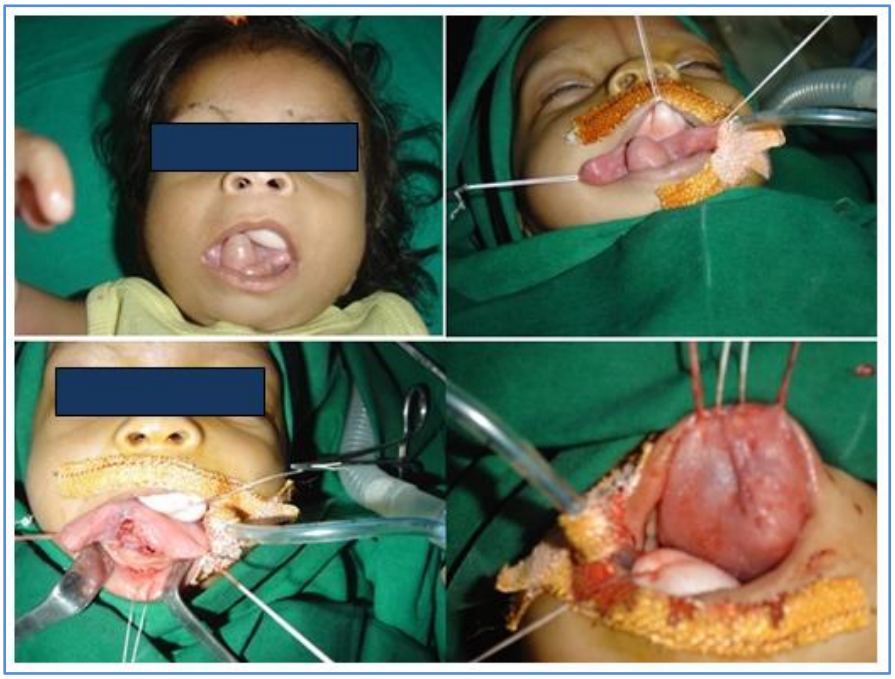

Fig. 1: various images of bifid tongue

Surgical correction of the defect taken under general anesthesia. First mucous cyst was excised. Tongue was reconstructed by refreshing the defects and sutured in layers. The palatal swelling is excised. Post-surgical healing was uneventful (Fig.2).

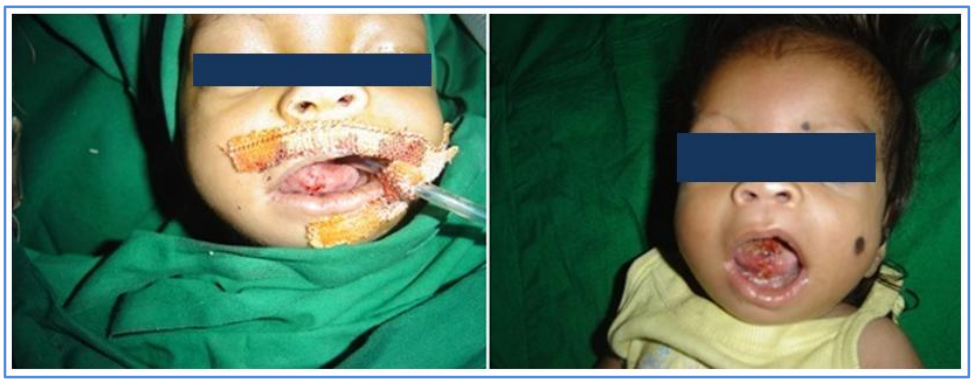

Fig. 2: Post-operative images

DISCUSSION: It is reported in the literature that median tongue clefts only to be associated with orofacial digital syndromes type I, II, IV and VI.[2,4,5] These syndromes are all associated with mandibular clefts, median lip, bilateral dislocation of knees, elbows, ankles, pulmonary hypoplasia and dysmorphic facial features including prominent forehead, depressed nasal bridge and widely spaced eyes were suggestive of Larsen-like syndrome type 1,[6,7,8] optic G BBB syndrome, oral facial

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digital syndrome type 1, Klippel-Feil anomaly. Bifid tongue has also been reported as a rare feature associated with infants of diabetic mother.[2,3,9]

Congenital lipomas of the fornix of the vestibule and of the tongue were reported in a 7 month old boy and 20 day old girl respectively. Congenital hairy polyps of the nasopharynx in association with cleft palate were diagnosed in two new born.[10] Oral hamartoma usually arises from the foramen cecum during embryonic development, which may explain why most LLH are in the midline of the tongue.[1]

The development of the tongue starts at the fourth week of intrauterine life in the floor of the primitive cavity from the first three or four branchial arches. Abnormal/ partial/ non-fusion of these arches may lead to congenital anomalies of tongue, including bifid tongue.[2] Our case was not associated with any syndrome or any other associated orofacial abnormalities or no history of post natal trauma, no genetic predisposition, no tongue piercing.

CONCLUSION: Bifid tongue can be described as cleft tongue/ diglossia, though many orofacial syndromes associated with it. Our case cannot be put in any well-defined syndrome and the palatal hamartoma with bifid tongue in a 5 month old infant is a rare case.

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