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Original Research Paper

Otorhinolaryngology

CASE SERIES OF BIFID TONGUE: AS AN ISOLATED ENTITY

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ABSTRACT Period of organogenesis is very precise duration, any factor affecting this period leads to congenital anomaly. Bifid tongue is one of the congenital anomalies mostly associated with syndromic cases like Opitz G/BBB syndrome, oral–facial– digital syndrome type I, but isolated bifid tongue can also be seen rarely. Here we describe three cases of isolated bifid tongue in absence of any intra oral or facial defect.

KEYWORDS : Bifid tongue, Congenital anomaly, Isolated syndrome, Opitz G/BBB syndrome.

INTRODUCTION

Tongue is having important function in human life. Any deformation of the tongue during organogenesis leads to many anomalies such as tongue tie, bifid tongue, microglossia or macroglossia. These anomalies are ultimately leading to speech impairment or cosmetic problems. Tongue development begins in fourth week of intra uterine life by one median swelling (tuberculum impar) and two lateral lingual swellings. These two lateral lingual swellings grow rapidly and cover median swelling to form anterior two-third of tongue [1]. Any factor that can affect this developmental process can leads to malformation. Bifid tongue can be seen as a part of any syndrome or isolated finding [2].

MATERIAL AND METHODS

This is a retrospective study carried out in the department of Otorhinolaryngology of a tertiary care centre of eastern Uttar Pradesh, India in a duration of two years.

Inclusion criteria

Patients of isolated bifid tongue, Patient willing for consent, Patient of any age group and gender

Exclusion Criteria

Patients with associated congenital anomaly, Patients not willing for consent

Case Report

Case 1

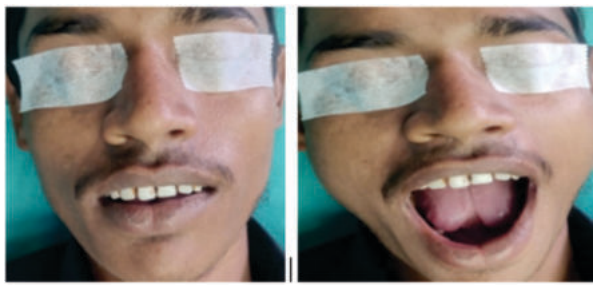
A young boy aged 21 years presented to our out-patient department with cosmetic deformity in the tongue. Patient had no functional impairment which led to delay in consultation. Patient was healthy without any significant antenatal or medical history. Rest of the oral cavity and facial examination is normal with no sign suggestive of congenital orofacial defect. After proper history including diabetes mellitus in family specially in mother, trauma, tongue piercing/surgery and general examination, he underwent routine investigations and was planned for surgical correction under local anesthesia. The edges of the defect were excised using sharp blade and wound was sutured in layers. The patient was instructed for proper oral hygiene and care. Post operative period was uneventful. In further follow up, tongue healed without any complication.

Case 2

A 17-year female visited ENT outdoor for cosmetic deformity of the tongue. She had no significant antenatal or medical history. Complete personal and family history, general examination was done for any congenital defect. Examination of face and oral cavity was done to rule out any congenital orofacial defect. After routine investigations, she was posted for surgical correction under local anesthesia. The edges of the defect were excised using sharp blade and wound was repaired in layers. Care was taken to ensure oral hygiene post-operatively. A good cosmesis of tongue was achieved post-operatively.

Case 3

A 16-year male patient reported to outpatient department with difficulty in speech and cosmetic problem due to tongue defect. Detailed history including diabetes mellitus in family, trauma, tongue piercing/surgery was taken followed by general examination which was unremarkable ruling out any congenital orofacial defect. Local examination including oral examination, nasal endoscopy and laryngoscopy was performed which was normal except a bifid defect in tip of the tongue. Routine investigations were done, and the case was planned for surgical correction under local anesthesia. The margins of the defect were freshened, and layered suturing was done. The patient was instructed for proper oral hygiene and post-op care which remained uneventful. In further follow up, tongue healed without any complication. Speech became normal in subsequent follow up without speech therapy.



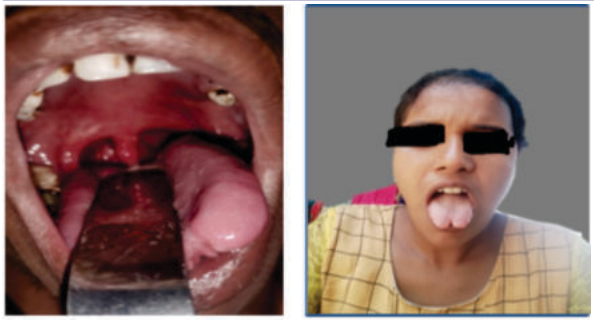


Fig.1 Various Preoperative Pictures of Bifid Tongue

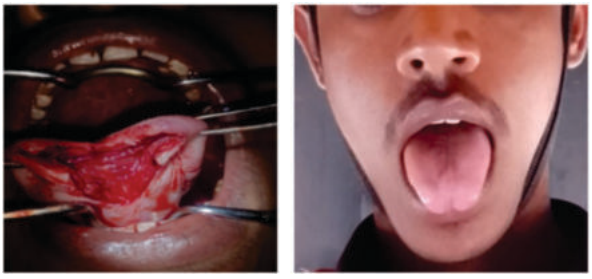


Fig.2 Intraoperative and Postoperative Pictures of Patient with Bifid Tongue

DISCUSSION

The first three or four brachial arches are the precursor of the tongue in primitive oral cavity during the fourth week of intra uterine life [1]. Any factor affecting the mesenchymal fusion during this period leads to congenital anomaly. Bifid tongue, in majority of cases is associated with other syndromes but can be as an isolated entity [2]. Fleming et al. reported that bifid tongue can occur as a complication of tongue piercing [3]. The frequency of congenital tongue malformations is aglossia, syndromic microglossia, macroglossia, accessory tongue, long tongue and bifid or cleft tongue in decreasing order of incidence. Parents of the syndromic cases of bifid tongue seek early medical attention but, in the cases, where bifid tongue is an isolated entity, this is neglected and presents later in the life [4]. Infants of the diabetic mother can also have bifid tongue as a rare feature [5]. Bifid tongue can be associated with many syndromes like Opitz G BBB syndrome, oral–facial– digital syndrome type I, Klippel–Feil anomaly and Larsen syndrome [6-9]. It can be associated with cleft palate, mandibular cleft, midline palate-mandibular bony fusion or cervical vertebrae [10]. Bartholdson et al described a case of cleft palate combined with bifid tongue in a baby boy [11].

CONCLUSION

Bifid tongues are due to the malformation of tongue during intra uterine life. Isolated bifid tongues are rare entity and attendants usually do not seek medical attention in early childhood which can result into speech abnormalities, dysphagia or merely a cosmetic defect. Surgical correction of the defect is mainstay of treatment.

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