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Non-Syndromic Bifid Tongue - A Case Report

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Abstract

Bifid tongue is a rare condition that can be syndromic or nonsyndromic and usually occurs in conjunction with other oral abnormalities. In oro-facial-digital syndrome and Tessier type 30 craniofacial cleft, bifid tongue has been frequently reported as an associated finding. Although rare, it has been found in conjunction with other syndromes and in nonsyndromic cases. A 51 year old female patient presented with a bifid tongue tip involving the anterior one-third of the tongue and found the patient unusually with no other intraoral abnormalities. The patient was in good health, his vital signs were within normal ranges, and he had no relevant family or medical background. Any other related congenital orofacial defects were ruled out by extra oral and intraoral analysis. No associated clinical features suggesting any syndrome was evident in this case. With this article, the patient clinical features on examination suggests that bifid tongue need not always be associated with a syndrome, nonsyndromic bifid tongue can also be found on ruling out possible syndromic association.

Keywords: Bifid Tongue; Cleft tongue; Developmental abnormality; Tongue; Case report

Introduction

During the fourth week of pregnancy, the tongue grows from a central swelling, the tuberculum impar on the pharyngeal surface, and two lateral lingual swellings that join this central structure. These lateral lingual structures quickly develop to cover the tuberculum impar and form the tongue's anterior two-thirds. First the tip of the tongue is separated longitudinally for a certain distance when this mechanism is disrupted, resulting in cleft tongue/bifid tongue [1]. Bifid tongue in the absence of other orofacial disorders is an unusual condition, and it normally gets much less attention than those seen in conjunction with certain syndromes [2]. Human development is a process that must be meticulously planned and executed. Even a minor ailment can lead to clinically significant consequences later on [3,4]. As seen in the case report presented here, ancient beliefs and practices continue to override/delay a patient's desire to seek treatment. Congenital bifid tongue has been linked to a number of orofacial anomalies [5]. A differential diagnosis must be made when a patient presents with a bifid tongue in order to arrive at a definitive diagnosis that will aid in identification, classification, management, and documentation. We present the patient's history, clinical features, and make differential diagnosis of syndromes associated with syndromes or non-syndromic and further aid in identifying cases which can be non-syndromic.

Case Report

A 51 year old Indian female patient, visited the clinic with a chief complaint of difficulty in speech from childhood. Patient revealed the history of consanguineous marriage. Her mother did not have any previous abortion or drug during pregnancy. Patient had no relevant medical and surgical history. Patient had no other difficulties other than aphasia. On physical examination at that time, she was noted to have obesity and hypertension. Her body mass index was 29kg/m^2. Lab testing revealed an abnormal glucose loading test and abnormal 3-hour glucose tolerance test. On clinical examination of the oral cavity revealed the

tongue was bifid at the tip which was slightly towards the right (Figure 1). In both the upper and lower vestibules, there were several frenal attachments. The upper frenii were hypertrophic and bilaterally clefted the upper alveolus. The clefts were found between the upper lateral incisors and canines, though the deciduous canines were found in the premaxillary segment. On the left side of the lower lip, there was a single mucous pit. Extra oral examination showed slanting palpebral fissures, alopecia, and a large tip to the nose, excessive facial hair, and dry coarse hair all over the scalp. Examination of the rest of the body revealed normal fingers and toes. Systemic examination did not reveal any abnormality. Our case defies categorization into any well-defined syndrome. There were no orofacial defects, hereditary predispositions, tongue piercings, or postnatal trauma histories found. Surgical correction of the defect was planned under local anaesthesia. The median parts of the defect were freshened and reconstructed by suturing the muscles in layers. The patient was not willing for surgical management.

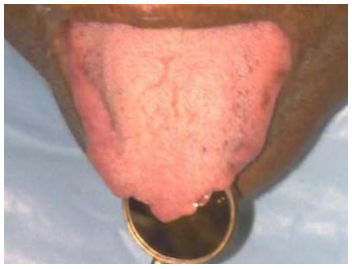

Figure 1: *Picture representing bifid tongue in anterior 2/3rd of the tongue.*

Discussion

The appearance of the median tongue bud at the end of the fourth embryonic week is the first sign of tongue formation. On either side of the median tongue bud, lateral lingual swellings, also known as distal tongue buds, appear shortly after. These swellings are caused by mesenchymal proliferation of the pharyngeal arch's first pair. The distal buds grow in size, merge in the midline, and form the tongue's anterior two-thirds, also known as the oral segment. The fusion is marked by a middle groove on the tongue called the median sulcus.

A tongue with a groove or split running lengthwise along the tip (glossoschissis) is known as a bifid or cleft tongue. It's caused by a lack of fusion between the distal tongue buds. A bifid tongue may be a one-of-a-kind condition, but it has also been linked to maternal diabetes. Two babies with a bifid tongue were born to diabetic mothers [6]. Only orofacial digital syndromes type I, II, IV, and VI have been linked to median tongue clefts in the literature. All of these syndromes are linked to clefts in the median lip and/or mandible, as well as digital variations. Some malformations discovered in those babies included a cleft palate and polydactyly. Various variants of the heterogeneous group of oral-facial-digital syndromes are believed to manifest as combined deformities of the palate and tongue [7]. Vandenhaute et al reported a child with the Pierre-Robin sequence, epignathus teratoma, and a bifid tongue died at the age of two months. In around 6% of cases, epignathus teratomas of the oropharyngeal region are associated with other malformations, such as a bifid tongue or even a bifid nose. Other syndromes with a bifid tongue have been identified, including short rib syndrome, short rib polydactyly syndrome, median cleft syndrome, and the Klippel-Feil anomaly [8].

The diagnostic criteria of any well-defined condition are not fully met in our case. While minor digital bone abnormalities can be missed on a propositus radiogram, the diagnosis of an oral-facial-digital syndrome is doubtful due to the lack of characteristic digital features [5]. Due to the absence of characteristic stigmata such as hypertelorism and a deep nasal root, it was also difficult to diagnose a median cleft syndrome [9]. The lack of fused cervical vertebrae ruled out the Klippel-Feil anomaly. Nonetheless, the Pierre-Robin series can be classified based on the combination of abnormalities found in our case. Micrognathia, a cleft palate, posterior retraction of the tongue, and neonatal breathing problems are the most common characteristics of this congenital malformation, which has a heterogeneous and largely unknown etiology [4].

Conclusion

Bifid tongue is a rare condition which is mostly encountered as a developmental abnormality. Non-syndromic bifid tongue is reported in this article which suggests that bifid tongue need not always be found associated with syndromes. Proper patient history and clinical examination must be ensured before misdiagnosing it as a syndromic.

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