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Prevalence of Bifid Tongue and Ankyloglossia in South Indian Population with an Emphasis on its Embryogenesis

Prevalencia de Lengua Bífida y Anquiloglosia en el Sur de la Población India con Énfasis en su Embriogénesis

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SUMMARY: Disturbance in the organogenesis of tongue might lead to some malformations like tongue tie, bifid tongue and hairy tongue. Severe degrees of these anomalies may cause speech impairment or periodontal defects. The present study was done on patients of the southern coastal belt of India during the past two years, on gross tongue anomalies. The results of the present study reveal that occurrence of tongue tie is 0.2% and bifid tongue is 0.3% in the southern coastal population. Since great majority of these oral anomalies have genetic basis the purpose of the present report is to highlight that these anomalies can exist without any familial background and also to suggest that environmental factor may play a role in the etiogenesis of these anomalies.

KEY WORDS: Bifid tongue; Inheritance; Prevalence tongue anomalies; Tongue tie.

INTRODUCTION

Since the early days of medical history tongue acts as an index marker in early diagnosis of some of the systemic diseases. Hippocrates, Galen and others considered the tongue as the barometer of health, emphasizing its diagnostic and prognostic importance (Bouquot & Gundlach, 1986). The tongue appears in embryos of approximately 4 weeks in the form of two lateral lingual swellings and one medial swelling, the tuberculam impar (Sadler, 2010). These 3 swelling originate from the first pharyngeal arch. Another median swelling, the copula or hypobranchial eminence is formed by mesoderm of the second, third, and part of the fourth branchial arch which gives rise to posterior part of the tongue (Sadler; Hamilton *et al.*, 1976). Besides relatively frequent tongue lesions, the prevalence of some rarely occurring conditions as reported earlier are bifid tongue 0.4%, ankylosed tongue 0.8%, microglossia (Mojarrad & Vaziri, 2008). These tongue lesions showed no association with sex or age. Ugar-Cankal *et al*. (2005) reported a higher prevalence for ankyloglossia, bifid tongue seen in boys than in girls in his study. Ankyloglossia was seen in 1.5% of boys and 1.1% in girls. Bifid tongue was 0.6% in boys and 0.2% in girls. The present study reports about the incidence of bifid tongue and ankyloglossia in south Indian population.

PATIENTS AND METHOD

The present study is based on the clinical investigations done during the past two years, on children having gross tongue anomalies like ankyloglossia & bifid tongue. The patients of the southern costal belt of India were selected for the study along with their familial history and genetic make up. Each of these patients was examined for genetic diseases.

RESULTS

Among 1800 patients Bifid Tongue (Fig. 1) was observed in 3 patients without any familial history. Ankyloglossia (Fig. 2) was observed as early as at the age of 6 weeks, and it was observed in 4 children, with clinical symptoms including not able to protrude the tongue, and

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dimpling of the tip of the tongue on any attempt of protrusive movement. All these patients did not exhibit diabetic history of the parents.

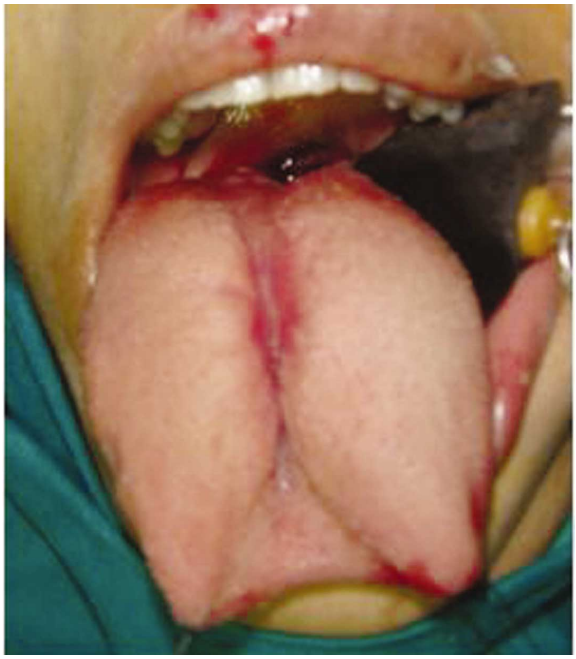

Fig. 1 Oral cavity showing bifid tongue.

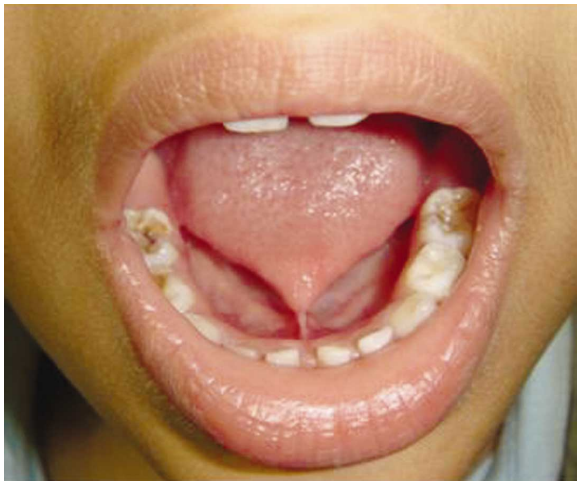

Fig. 1 Oral cavity showing bifid ankyloglossia.

DISCUSSION

Disturbance in the organogenesis of tongue might lead to some malformations like tongue tie, bifid tongue and hairy tongue. Severe degrees of these anomalies may cause speech impairment or periodontal defects. According to Ugar-Cankal *et al*. commonly found tongue anomalies are ankyloglossia (1.3%), fissured tongue (0.9%), bifid tongue (0.4%), hairy tongue (0.2%) and macroglossia (0.2%), lingual thyroid nodule (0.1%). Infants born to diabetic mothers are well documented to have a higher rate of congenital malformations (James *et al*., 2007; Bhuiyan *et al.*, 2009).

Bifid tongue. Bifid tongue occurs due to failure of fusion of above mentioned branchial arches. Prevalence of it has been reported with babies of diabetic mother. James *et al*. describe a male infant born to a diabetic mother who in addition to other typical congenital abnormalities was born with an impressive bifid tongue. On the contrary, Bhuiyan et al. report about a 2-day old baby with dermoid over hard palate associated with bifid tongue, born to a non-diabetic mother. Orhan *et al*. (2008) describe a 24 wk old aborted female fetus having bifid tongue as autosomal dominant & autosomal recessive patterns of inheritance. Mihci *et al*. (2007) explain a 9-month old girl having facial dysmorphology in addition to cleft palate and bifid tongue with multiple masses on the tongue, transmitted as a X linked dominant condition, which has to be ruled out with the oral facial digital syndrome Type I. Orhan *et al*. report about 24-week old aborted female fetus having lethal type Larsen-like syndrome associated with bifid tongue for the first time suggesting that every patient with Larsen-like syndrome should be examined carefully for the possibility of this abnormalities. Majority of the above reports suggest that bifid tongue occurs in addition to some other anomalies. However, in the present case bifid tongue was not associated with any other deformity.

Ankyloglossia. Commonly known as tongue tie, is a congenital oral anomaly which may decrease mobility of the tongue tip and is caused by an unusually short, thick lingual frenulum, a membrane connecting the underside of the tongue to the floor of the mouth (Horton *et al.*, 1969). Ankyloglossia varies in degree of severity from mild cases characterized by mucous membrane bands to complete ankyloglossia whereby the tongue is tethered to the floor of the mouth (Sadler). Along the ventral and sides of the rudimentary tongue develops an endodermal alveolo-lingual sulcus which gradually separates the tongue from the floor of the mouth. The pathogenesis of ankyloglossia is not known. It can be part of certain rare syndromes such as X linked cleft palate (Moore *et al.*, 1987) and van der Woude syndrome (Burdick *et al.*, 1987). Maternal cocaine use is reported to increase the risk of ankyloglossia to more than threefold (Harris *et al*., 1992). Morowati *et al.* (2010) reported a family with isolated ankyloglossia inherited as an autosomal dominant or autosomal recessive trait having prevalence of 4.5% with abnormally short lingual frenulum common in males. Khozeimeh & Rasti (2006) reported partial ankyloglossia of 5% in school children in Borazjan, Iran. A study in Nigeria by Sawyer *et al.* (1984) reported ankyloglossia as 0.2% incidence and in Saudi Arabia as 0.1% as by Salem *et al*. (1987). The present report reveals that among South Indian population the occurrence of tongue tie is relatively rare with an incidence of 0.2%.

CONCLUSION. It is important to look for the genetic etiology of all diseases and anomalies to have specific treatments and prevent them. This study has demonstrated that in South Indian population bifid tongue and ankyloglossia can occur without any genetic inheritance which suggests that occurrence of these anomalies vary in different races and different environmental conditions. The identification of the factors causing these anomalies might reveal novel information about craniofacial embryogenesis and the pathogenesis of this disorder.

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RESUMEN: La alteración en la organogénesis de la lengua puede dar lugar a algunas malformaciones como anquiloglosia, lengua bífida y lengua vellosa. Grados severos de estas anomalías puede provocar un trastorno del habla o defectos periodontales. El presente estudio se realizó, durante los últimos dos años, en pacientes de la franja costera del Sur de la India con anomalías graves en la lengua. Los resultados del estudio revelaron que, en la población costera del sur, la incidencia de anquiloglosia era de 0,2% y de lengua bífida de 0,3% . Dado que la gran mayoría de estas anomalías orales tienen base genética, el propósito del presente informe fue poner de relieve que estas anomalías pueden existir sin ningún tipo de antecedentes familiares y también sugerir que los factores ambientales podrían jugar un papel en el etiogenesis de estas anomalías.

PALABRAS CLAVE: Lengua bífida; Herencia; Prevalencia de las anomalías de la lengua; Anquiloglosia.

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