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Case Report

Ankyloglossia, A Case Report: Untying the Tongue Tie in the Family Tree

Sphoorthi Basavannaiah*

Associate Professor, Department of ENT, Subbaiah Institute of Medical Sciences, India

***Corresponding Author:** Sphoorthi Basavannaiah, Associate Professor, Department of ENT, Subbaiah Institute of Medical Sciences, India

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Abstract

Ankyloglossia or 'Tongue-tie' is a congenital oral anomaly characterized by the persistence of the lingual frenulum as an anatomical abnormality. It has a prevalence of 0.1 – 10.7 % as reported in the literature with affection more towards males. I hereby present a case report to have come across an Isolated Ankyloglossia in a family of 4 members, in whom 3 are affected with this condition.

Keywords: Ankyloglossia/Tongue tie; inheritance; familial; congenital; syndrome

Introduction

Ankyloglossia is a congenital anomaly characterized by unusually short lingual frenulum. It can result in varying degree of decreased tongue mobility leading to feeding and speech difficulty and also debatable social issue due to hampered tongue protrusion [1,2]. The pathogenesis of Ankyloglossia is anonymous and its management has been divisive. Though, relatively common it has a prevalence of 0.1 – 10.7 % as reported in the literature with affection more towards males (3:1) [3,4]. In this case report, a family with isolated Ankyloglossia is mentioned in which 3 individuals over 2 generations have Ankyloglossia, inherited as an autosomal dominant trait. As exact pathogenesis is unknown and also there is limited accessibility to case reports, studies regarding genetic predilection of tongue tie. Here, I present this as an assenting case report explaining genetic inheritance pattern of tongue tie.

Case Report

A mother brought in her 2 boys aged 6 and 9 years to the ENT outpatient department who presented with slurred speech and inability to protrude the tongue. Following clinical examination, both were diagnosed to have Ankyloglossia (Class III as per Kotlow [5] assessment). Family history revealed the presence of tongue tie in their father as well. Hence, 2 generations in the family were affected. There was no similar history in other family members in both paternal as well as maternal side. The boys were treated at our department and the tongue tie was released (Frenotomy). The father was apprehensive to undergo release of the tongue tie and wished to undergo release after going through his children's response. Postoperatively, patients were advised to start touching the tip of the tongue to the palate (which initially they were unable to do) and make the sound "tataaatataaa" and usage of words to articulate/ speak where the tongue stresses and touches the palate such that there would be more tissue expansion and extensibility.

Discussion

Ankyloglossia, a congenital anomaly is said to have an unusually short lingual frenulum. There is no constant description or classifying system to describe this condition. This clinical disorder varies from lack of medical implication to erratic ample consequence where the ventral part of tongue is fused to the floor of mouth [1,4]. Tongue tie can be considered a relatively common anomaly with a prevalence ranging from 0.1 -10.7%. The Online Mendelian Inheritance in Man (OMIM) has given identification number for ankyloglossia as 106280 [1]. In neonates, the prevalence is 1.72% - 10.7%, while in adolescents or adults it ranges from 0.1% - 2.08%. For unknown reasons the abnormality seems to be more common in males, with male to female ratio 3:1. The exact pathological mechanism of ankyloglossia remains unclear and its conclusive hereditary nature is yet to be elucidated [1,3,4]. Ankyloglossia, though being congenital can also be a part of certain rare craniofacial disorders and syndromes such as X- linked cleft palate, Van der Woude syndrome, Ehler Danlos syndrome, Beckwith Wiedemann syndrome, Smith –Lemli-Opitz's syndrome, Orofacial

digital syndrome and Simpson-Golabi-Behmel syndrome [2,6]. Most of the times, Ankyloglossia is seen as an isolated finding in an otherwise normal child. There is a 3- fold increased risk reported to Ankyloglossia when on maternal cocaine use [7]. Tongue tie may result in varying degree of decreased tongue mobility. It has been proposed to cause breast feeding difficulties such as sore nipples, poor infant weight gain, early weaning, speech disorders like impaired articulation, limited tongue protrusion, difficulty in speech, problems with deglutition, chewing food, dentition, difficulty to wear a denture at various stages of life, maintaining oral hygiene, stress and social issues related to limited function of the tongue [1,2,3]. Management of ankyloglossia has been controversial. For this condition treatment options include surgical procedures as Frenotomy, Frenectomy, Frenuloplasty Lasers and Electrosurgery though its spontaneous resolution is also possible in some cases. No widely accepted criteria have been established for the surgical indications and the selection of surgical procedures. There is no common view regarding the hints, timing or method of surgical repair for ankyloglossia [1,2,8].

In the above-mentioned case the affected father has passed this condition onto the affected off springs. This appears to be inherited autosomal dominant trait [3,4,8,9]. As per various case reports that are mentioned below, despite there are very limited studies regarding familial inheritance of this condition: As per study by Morawati et al . [4], 5 individuals were affected in 5 generations with inheritance of autosomal dominant or autosomal recessive trait. According to study by Keizer et al [9], a Dutch family in which 13 persons in 3 generations were affected having a male-to-male transmission. The study by Klockars et al. [8], 9 individuals spread over 4 generations were affected with inheritance of autosomal dominant trait. As per study by Pakanati et al. [3], similar findings as Klockars study were mentioned. Except for tongue tie, defect in articulation, clarity of speech, and difficulty in chewing food, I did not find any other symptoms or anomalies in the children in this case report. Previous literature suggests cases with fibrous bands associated with congenital abnormality such as anencephaly, tracheo- oesophageal fistula or cleft palate. LGR5 gene [Orphan G-protein – coupled receptor gene] [10] is found to be associated with neonatal lethality and ankyloglossia in mice and TB X 22 gene [T-box transcription factor gene] [11] which is mutated in X- linked cleft palate and ankyloglossia.

Conclusion

Tongue – tie is a fairly innocuous condition, and its treatment is simple and safe when treated at an optimum time. It not just has a part to play in articulation, deglutition and mastication but tends to avoid the glitches of social taboos and issues and builds in the lack of self-confidence. The hereditary incidence of ankyloglossia is erratic and infrequent. To understand the genetic aspect of this condition more such studies should be through and reconnoitred. As per literature, there is limited information provided on the pedigree inheritance of this condition. Hence, identification of defective gene/s causing this condition might reveal innovative material about craniofacial embryogenesis and pathogenesis of this disorder.

References

1. Devasya A, Sarpangala M (2017) Familial Ankyloglossia-A Rare Case Report. Journal of Clinical and Diagnostic Research 11(2): 3-4.
2. Jangid K, Alexander AJ, Jayakumar ND (2015) Ankyloglossia with Cleft lip: A rare case report. Journal of Indian Society of Periodontology 19(6): 690-693.
3. Pakanati SSR, Anchery VA, Moidu F (2014) Familial ankyloglossia [tongue – tie] – a rare case report., Journal of Evolution of Medical and Dental Sciences 3(2): 303-305.
4. Morowati S, Yasini M, Ranjbar R (2010) Familial Ankyloglossia (Tongue-tie): A Case Report Acta Medica Iranica 48(2): 123-124.
5. Kotlow LA (1999) Ankyloglossia (tongue-tie): A diagnostic and treatment quandary. Quintessence Int 30: 259-262.
6. Burdick AB, Ma LA, Dai ZH (1987) Van der Woude syndrome in two families in China. J Craniofac Genet Dev Biol 7(4): 413-418.
7. Harris EF, Friend GW, Tolley EA (1992) Enhanced prevalence of ankyloglossia with maternal cocaine use. Cleft Palate Craniofac J 29(1): 72-76.
8. Klockars T (2007) Familial ankyloglossia (Tongue-tie) Int J Pediatr Otorhinolaryngol 71: 1321-1324.
9. Keizer DP (1952) Dominantly hereditary ankyloglossia. Ned Tijdschr Geneeskd 96: 2203-2205.
10. Morita H, Mazerbourg S, Bouley DM (2004) Neonatal lethality of LGR5 null mice is associated with ankyloglossia and gastrointestinal distension. Mol Cell Biol 24(22): 9736-9743.
11. Braybrook C, Doudney K, Marçano AC (2001) The T-box transcription factor gene TBX22 is mutated in X-linked cleft palate and ankyloglossia. Nat Genet 29(2):179-183.

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