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Familial Ankyloglossia -A Rare Report of three Cases in a Family

ASHWIN DEVASYA[1], MYTHRI SARPANGALA[2]

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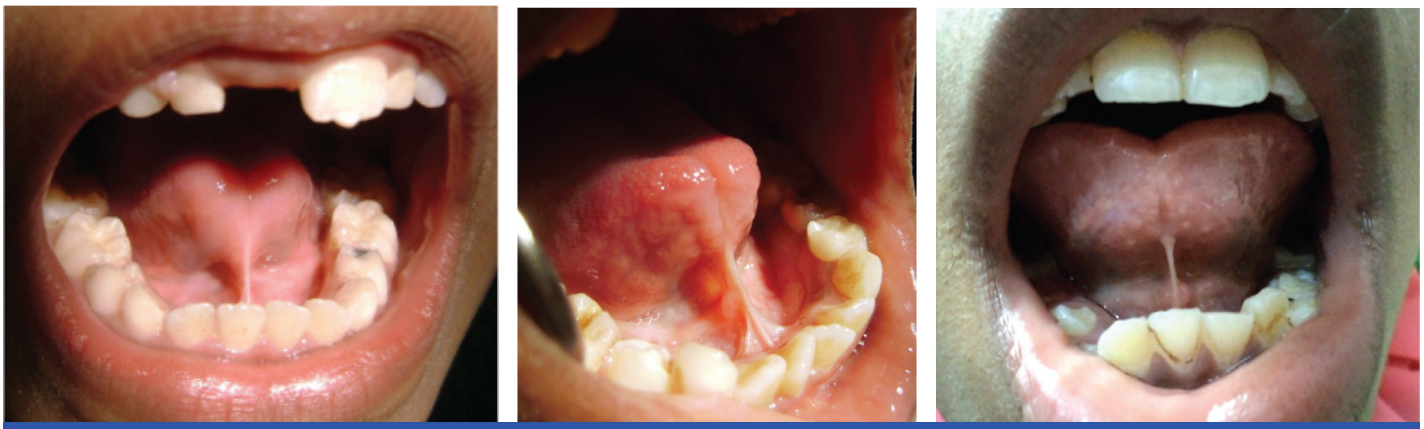
Ankyloglossia, commonly known as 'tongue-tie', is a congenital oral anomaly characterized by the persistence of the lingual frenulum as an anatomical abnormality [1]. The Online Mendelian Inheritance in Man (OMIM) has given identification number for ankyloglossia as 106280 [2].

The prevalence reported in the literatures varies from 0.1% to 10.7%. In neonates the prevalence is 1.72% to 10.7%, while in adolescents or adults it ranges from 0.1% to 2.08% [3]. It affects males more commonly, in the ratio 3:1 [3].

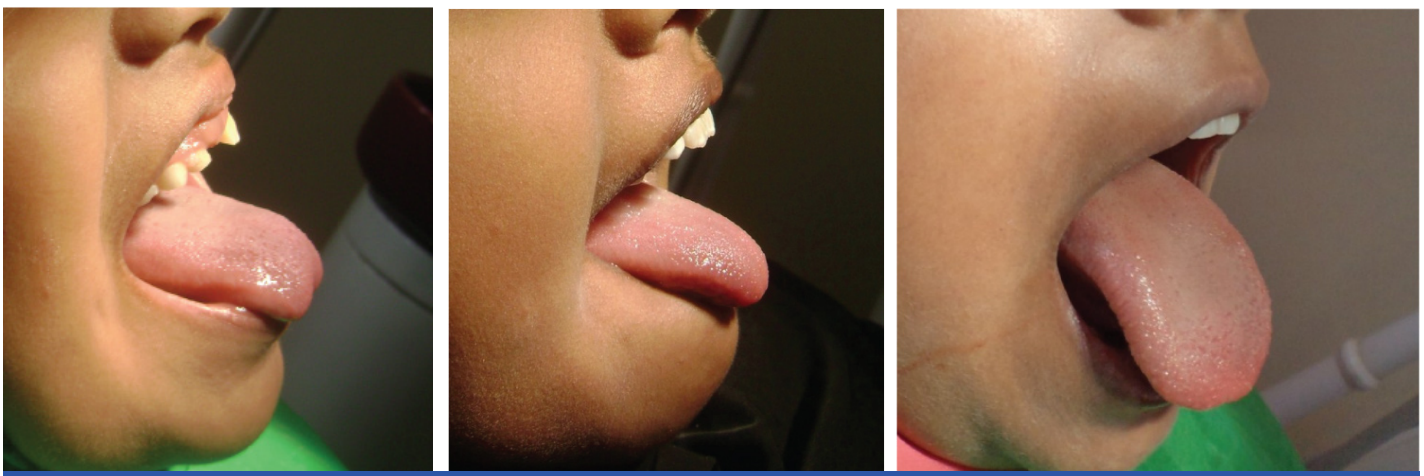
There are very few cases of familial ankyloglossia reported till now. In this report, we have described a family affected with ankyloglossia in two generations inherited as an autosomal dominant or recessive trait. Five individuals in two generations were affected out of which three are described here. Most importantly, in this family females were affected more than males.

A parent came to our hospital with a complaint that his nine-year-old daughter has a difficulty in pronouncing few words. On intraoral examination it was found that the individual had ankyloglossia or tongue tie and was classified as Class 2 [Table/Fig-1], based on Kotlow's assessment [4]. Parents were explained the reason, problems caused by the condition and the need for immediate treatment.

Interestingly, father also mentioned that his wife (the mother) and two of his daughters and a son also have the similar problem, son



[Table/Fig-1]: Case 1: A nine-year-old female diagnosed as Class 2 ankyloglossia based on Kotlows's assessment.
[Table/Fig-2]: Case 2: An eight-year-old female sibling with class 3 ankyloglossia based on Kotlows's assessment.
[Table/Fig-3]: Case 3: An 11-year-old female sibling with Class 2 ankyloglossia based on Kotlows's assessment.



[Table/Fig-4]: Case 1: Follow up after a week showing good healing and extensibility of tongue.
[Table/Fig-5]: Case 2: Follow up after a week showing good healing and extensibility of tongue.
[Table/Fig-6]: Case 3: Follow up after a week showing good healing and extensibility of tongue.

being their eldest child. There was no other history of tongue tie in their family. The other two daughters eight-year-and 11-year-old were also examined and we found that they have Class 3 [Table/Fig-2] and Class 2 [Table/Fig-3] type ankyloglossia respectively, based on Kotlow's assessment.

The son and the mother did not appear for the examination or the treatment. But based on the details given by the father we just have mentioned the incidence in this report which is not confirmative.

All the three children underwent frenectomy procedures using single haemostat method and were advised tongue exercises after a week. Follow up after a week showed remarkable healing of tissues [Table/Fig-4-6] and after six months there was reduction in pronunciation problems of the children.

DISCUSSION

Ankyloglossia is congenital and sometimes associated with various syndromes like Ehler Danlos syndrome, Beckwith Wiedemann syndrome, X-linked cleft palate and orofacial digital syndrome [5]. Ankyloglossia causes many problems like difficulty in breast feeding, difficulties in speech, oral hygiene, chewing food, stress and difficulty to wear a denture, in various stages of life [5]. For the purpose of treatment, there are various options available such as frenotomy, frenectomy, lasers and electrosurgery [4].

Familial ankyloglossia is a very rare condition. It has been reported that mutation of T-box transcripition factor (TBX22) gene during palatogenesis causes ankyloglossia [6].

Ankyloglossia can also be a part of X-linked cleft palate caused by mutations in the TBX22 gene. TBX22 is the sole phenotype in 4% of affected males and in 45% of carrier females. But in our cases, female predominance was more which seems to be rare [6].

CONCLUSION

Tongue tie is not a harmful condition and treating it at optimum time is safe and helpful in correction of problems. The familial occurrence of ankyloglossia is rare and uncommon. To understand the genetic aspect of it more studies should be done and explored.

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PARTICULARS OF CONTRIBUTORS:
1. Senior Lecturer, Department of Paedodontics and Preventive Dentistry, Kannur Dental College, Anjarakkandy, Kannur, Kerala, India.
2. Senior Lecturer, Department of Periodontology, Kannur Dental College, Anjarakkandy, Kannur, Kerala, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Ashwin Devasya,
Bhat's Multi Specialty Dental Clinic, Oberle Complex, Above Kottodal Hardwares, Badiyadka Road, Kumbla-671321, Kasargod, Kerala, India.
E-mail: ashwindkumbla@gmail.com

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