

ANKYLOGLOSSIA –A REVIEW AND CASE REPORT**^{1*}Dr. Manish Ashtankar, ²Dr. Mala Dixit Baburaj, ³Dr. Lashika Tambe and ⁴Dr Shraddha Kode**¹MDS Department of Periodontics & Implantology, Nair Hospital and Dental College, Mumbai.²Professor and HOD, Department of Periodontics & Implantology, Nair Hospital and Dental College, Mumbai.³III MDS Student, Department of Periodontics & Implantology, Nair Hospital and Dental College, Mumbai.⁴II MDS Student, Department of Periodontics & Implantology, Nair Hospital and Dental College, Mumbai.**Corresponding Author: Dr. Manish Ashtankar**

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ABSTRACT

Ankyloglossia, also known as tongue-tie, is a congenital oral anomaly that 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. 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. We have reported here a 22 year old male with sever ankyloglossia treated with frenectomy and speech therapy.

KEYWORDS: Ankyloglossia, frenectomy.**INTRODUCTION**

Etymologically, “ankyloglossia” originates from the Greek words “agkilos” (curved) and “glossa” (tongue). The frenulum of tongue or tongue web (also lingual frenulum or frenulum linguae) is a small fold of mucous membrane extending from the floor of the mouth to the midline of the underside of the tongue.

Definition

1. Wallace^[1] defined tongue-tie as “a condition in which the tip of the tongue cannot be protruded beyond the lower incisor teeth because of a short frenulum linguae, often containing scar tissue.”
2. As defined by the International Affiliation of Tongue-tie Professionals -The Embryologic remnant of the tissue in the midline of the undersurface of the tongue and the floor of the mouth.^[2]

Development

The tongue starts to develop at about 4 weeks. The tongue originates from the first, second, and third pharyngeal arches which induce the migration of muscles from the occipital myotomes. A U-shaped sulcus develops in front of and on both sides of the oral part of the tongue. This allows the tongue to be free and highly mobile, except at the region of the lingual frenulum, where it remains attached. Disturbances during this stage cause tongue tie or ankyloglossia.^[3]

Anatomy

The thin strip of tissue that runs vertically from the floor of the mouth to the undersurface of the tongue is called

the lingual frenulum. On either side of the midline, a ridge of fimbriated mucosa extends forward and medially towards the tip of the tongue. The aberrant lingual frenum consists of genioglossal muscle fibers.^[4]

The base of the frenulum contains a "V" shaped hump of tissue in the floor of the mouth which houses a series of salivary gland ducts. The two largest ducts are in the center just in front of the attachment of the lingual frenulum and are called Wharton's Ducts. They empty the submandibular (submaxillary) and sublingual salivary glands. These ducts can be quite active in some persons, and upon occasion, a considerable amount of saliva may erupt from them while talking, eating, yawning, or cleaning the teeth in a process known as gleeeking. The sublingual saliva glands empty through a series of tiny ducts in the tissue on either side of Wharton's ducts. The tongue is attached to the floor of the oral cavity by the frenulum.

Superficial veins run through the base of the frenulum known as varicosities.

PROBLEMS

- Speech and mastication: The most common difficulty is in the production of the “s” sound off the incisal edge of the lower incisor instead of behind the incisive papilla. The difficulties in articulation are evident for consonants and sound like “s, z, t, d, l, j, zh, ch, th, dg”¹⁷ and it is especially difficult to roll an “r”.
- Breastfeeding: tongue tie does not aid infant to latch

onto its mother and make feeding difficult. It also affects psychological bonding between mother and child. Mothers may experience nipple pain and/or sore nipples due to the excessive suckling pressure by the infant.

- Prosthesis construction: In the edentulous mouth even a well-compensated ankyloglossia may require surgical correction prior to the construction of full dentures.
- Inability to raise the tongue to the roof of the mouth may prevent the development of an adult swallow and encourage the continuation of the infantile swallow, which may lead to an open bite.
- Gingival recession on the lingual surfaces leads to the periodontal problem.
- Malocclusion: Mandibular prognathism and maxillary hypo development due to the low position and the forward and downward pressure applied.
- General consequences of ankyloglossia include social embarrassment due to disharmony of the speech, which may build up a complex within the child. Suckling inability due to improper seal leads to poor infant weight gain and early weaning.

Prevalence

Tongue tie can be considered a relatively common anomaly with a prevalence of <1–10.7%. The prevalence is also higher in studies investigating neonates (1.72% to 10.7%) than in studies investigating children, adolescents, or adults (0.1% to 2.08%).

A descriptive analysis of the prevalence of tongue lesions by Patil *et al.* in 2013 showed 21 patients out of 4926 Indian patients were affected with ankyloglossia, which accounts for 3.5% of the tongue lesions.^[18] For unknown reasons, the abnormality seems to be more common in males with a male to female ratio of 2.5:1.05.

Genetics

Appearance Score

Appearance of tongue when lifted	Elasticity of frenulum
2: Round or square	2: Very elastic
1: Slight cleft in tip apparent	1: Moderately elastic
0: Heart- or V-shaped	0: Little or no elasticity

Length of lingual frenulum when tongue	Attachment of lingual frenulum to tongue
2: > 1 cm	2: Posterior to tip
1: 1 cm	1: At tip
0: <1 cm	0: Notched tip

Attachment of lingual frenulum to inferior alveolar ridge
2: Attached to floor of mouth or well below ridge
1: Attached just below ridge
0: Attached at ridge

Perfect score = 10

Score is <8 = Ankyloglossia-indicated for frenectomy.

Most cases of ankyloglossia are sporadic, mutations in the T box transcription factor TBX22 may lead to heritable (X linked) ankyloglossia with or without cleft lip, cleft palate and/or hypodontia.^[6] Reports suggest that there is a genetic basis for the microvariation in the attachment of genioglossus muscle due to the mutations present in the TBX22 gene. In case of cleft palate with ankyloglossia (CPX) patients, the TBX22 expression is seen in early human development, where the expression is found in the palatal shelves and is highest prior to elevation to a horizontal position above the tongue.

TBX22 mRNA is also detected in the base of the tongue in the region of the frenulum that corresponds to the ankyloglossia seen in CPX patients.^[7]

A case report on familial ankyloglossia by Morowati *et al.* in 2010 reported a family of five generations in which five individuals had ankyloglossia inherited as an autosomal dominant or recessive trait.^[8]

Syndromes

Ankyloglossia was also found associated in cases with some rare syndromes such as X linked cleft palate syndrome, Kindler syndrome, Van der Woude syndrome, and Opitz syndrome.^[9]

Diagnostic criteria

Kotlow's assessment as follows

Class I: Mild ankyloglossia: 12 to 16 mm,
 Class II: Moderate ankyloglossia: 8 to 11 mm,
 Class III: Severe ankyloglossia: 3 to 7 mm,
 Class IV: Complete ankyloglossia: Less than 3 mm.^[10]

Hazelbaker Assessment for Lingual Frenulum Function^[11]

It measure 2 scores.

2. Function score

Lateralization	Lift of tongue	Extension of tongue
2: Complete	2: Tip to mid-mouth	2: Tip over lower lip
1: Body of tongue but not tongue tip	1: Only edges to mid-mouth	1: Tip over lower gum only
0: None	0: Tip stays at lower alveolar ridge or rises to mid-mouth only with jaw closure.	0: Neither of the above, or anterior or mid tongue humps

Spread of anterior tongue	Cupping	Peristalsis
2: Complete	2: Entire edge, firm cup	2: Complete, anterior to posterior
1: Moderate or partial	1: Side edges only, moderate cup	1: Partial, originating posterior to tip
0: Little or none	0: Poor or no cup	0: None or reverse motion

Snapback
2: None
1: Periodic
0: Frequent or with each suck

Perfect function score= 14.

Score of <11 =Ankyloglossia- indicated for frenectomy.

MANAGEMENT

1. Frenotomy is a simple cutting of the frenulum.
 2. Frenuloplasty: the surgical alteration of the frenulum can be performed in mild Class 1 (Kotlow's classification) cases.
 3. Frenectomy: the complete removal of the frenum and its attachment to the underlying structure has to be performed in Class II, III, IV cases, that is, moderate, severe and complete ankyloglossia.
- All these procedures can be performed using conventional scalpels, electrocautery or lasers.
4. After surgical correction, speech therapy for speech modulation.

CASE REPORT

A 22-year-old male was reported in the department of Periodontics with a chief complaint of pain and food lodgement in lower left third molar. After completion of treatment and follow up after 7days we asked the patient about his speech problem as he was not able to pronounce some words properly. The patient explained his speech problems severity as social embarrassment, lack of confidence. Further, he explained few months before he felt to get a job because of speech problem. He gave a history of discussion of the same problem with a family physician but did not get the solution. We explained him tongue tie as a reason for his speech and same can get treated by surgery. The patient was reluctant to accept the treatment as his experience with family physician. Finally, after proper counseling, he gave consent to carry lingual frenectomy procedure.

Clinical Examination

General physical and ENT examination was normal. On intraoral examination, the patient had ankyloglossia (tongue-tie) and was classified as Class IV by Kotlow's assessment [Figure 1] and was not able to protrude the tongue even up to the lower incisor and lip.



Fig. 1: Pre-operative view Class IV: Severe.

There were no malocclusion and recession present lingual to mandibular incisors. Caries was seen with 46 and temporary restoration with 37.

The patient was undertaken for a frenectomy procedure under local anesthesia with 2% lignocaine hydrochloride and 1:2, 00,000 adrenaline by using a scalpel method; first a curved hemostat was inserted into the bottom of the lingual frenum at the depth of the vestibule and clamped into position followed by giving two incisions at the superior and the inferior aspect of the hemostat. The intervening frenum was removed and left with a diamond-shaped wound. Then further dissection carried out with the help of the same hemostat to release the muscle fibers so as to achieve a good tension-free closure of the wound edges [Figures 2 and 3]. the wound edges were approximated with (4-0) black braided silk sutures [Figure 4] for the tissues to heal by primary intention thereby minimizing the scar tissue formation.



Fig.2: Hemostat in position before excision of frenum.

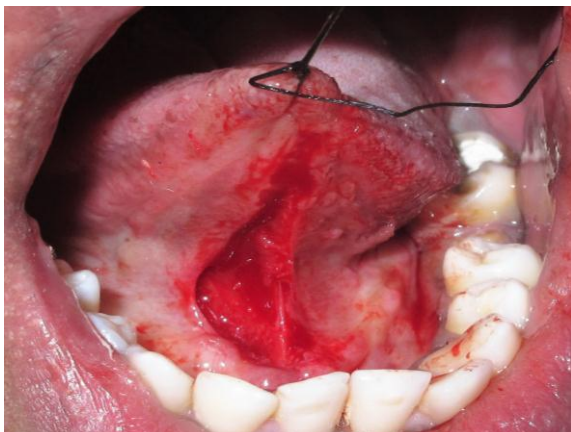


Fig.3: Frenum excision done.

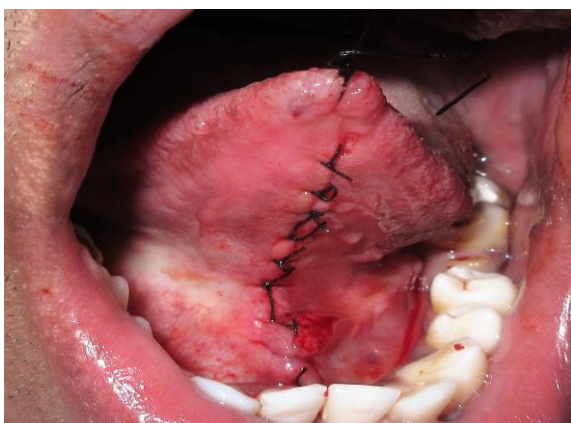


Fig.4: Suture Placed.

Antibiotic Cap. Amoxicillin (500 mg) thrice a day for 3 days and non-steroidal anti-inflammatory drug Tab. Diclofenac (50 mg) thrice a day for 3 days was prescribed to prevent post-operative infection and pain. The postoperative healing was uneventful with no delayed hemorrhage.

Sutures were removed after 1 week [Figure 5] which showed no scar tissue formation.

The patient was then sent for speech therapy sessions as the speech was his primary concern. After a follow-up of 9 months, the tongue showed good healing, protrusion

several mm beyond the lower lip [Figure 6], and normal speech.



Fig.5: Post-operative view after 1 week

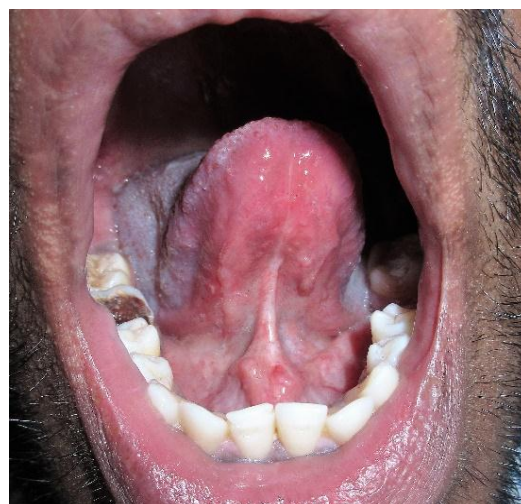


Fig.6: Post-operative view after 9 months

DISCUSSION

The term free-tongue is defined as the length of tongue from the insertion of the lingual frenum into the base of the tongue to the tip of the tongue. Clinically acceptable, normal range of free tongue is greater than 16 mm.^[12]

A normal range of motion of the tongue is indicated by the following criteria: The tip of the tongue should be able to protrude outside the mouth; without clefting, the tip of the tongue should be able to sweep the upper and lower lips easily; without straining, when the tongue is retracted, it should not blanch the tissues lingual to the anterior teeth; and the lingual frenum should not create a diastema between the mandibular central incisors. Ankyloglossia limits the tongue's range of motion.^[12]

Class III and IV tongue-tie category should be given special consideration because they severely restrict the tongue's movement.

It has been hypothesized that ankyloglossia can contribute to malocclusion. Limited mobility of tongue due to tongue tie keeps it at low position. This pressurizes mandible to grow in downward and forward direction

leads mandibular prognathism. There here is limited evidence to prove this hypothesis, so more clinical control studies required to correlate ankyloglossia as a cofactor with malocclusion.^[12]

The absolute pathogenesis of ankyloglossia remains unknown.^[14] While most cases of ankyloglossia are sporadic, mutations in the T-box transcription factor TBX22 may lead to heritable (X-linked) ankyloglossia with or without cleft lip, cleft palate and/or hypodontia.^[15]

Another case report in 1952 describes a Dutch family in which 13 persons in three generations had ankyloglossia and there were many instances of male to male transmissions.^[16]

In this case, report patient was having class IV ankyloglossia and he faced a lot of consequences of it. Consideration of such case for surgical management immediately is important. Frenectomy is defined as complete excision, i.e., removal of the whole frenulum. Frenectomy as surgical management can be done by scalpel, electrocautery and laser. The laser has an advantage over other two methods as less pain, clear surgical field and comfort of the patient but it is not cost effective.

Any of these methods does not influence healing drastically. There is no sufficient evidence in the literature concerning surgical treatment options for ankyloglossia to favor any one of the three main techniques.^[13]

In vitro studies have shown that healing does not show any considerable difference histologically when the three procedures are carried out on rats. Skeletal muscle fibers were better organized and dynamics of healing process were more rapid in scalpel group. Thus, though the approaches to the problem of not using the traditional scalpel have merited further improvements can still be made considering the healing aspect. Healing was complete by first post-operative month.^[9]

In some cases surgical management alone is not sufficient to improve speech of the patient. Speech therapy must include exercise for tongue such as oral kinesthesia (ability to feel the part and how they are moving) and DDK (diadochokinesis-ability to perform rapid, alternating movements) without which no significant improvement in speech will be achieved.^[9]

CONCLUSION

To conclude it is important to have awareness about ankyloglossia to all medical and dental specialists as it can affect individual's life socially and psychologically. Severe or complete ankyloglossia can cause problems like breastfeeding in infant, affect its growth, sleep apnea, speech problem, social and psychological trauma. So, diagnosis at an early age and its surgical

management followed by speech therapy can improve individual's life.

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