**Case Series** 

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# Severe Ankyloglossia and its Surgical Management: Case Report

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## Abstract

**Introduction:** Tongue frenulum is a band of mucosal tissue attaching the lingual bottom to the mouth's floor and mandibular arch. Most frenum abnormalities occur in the lingual site. The Ankyloglossia is one of these frenulum anomalies showing different classifications. Ankyloglossia can be silent or may have several symptoms. This condition may disturb skeletal development, affect oro facial functions and also may induce difficulties in oral hygiene.

**Method:** Two cases of severe ankyloglossia treated with conventional surgical correction by frenectomy procedure expressing immediate change in lingual posture and function.

**Results:** The reevaluation shows stability of the results and significant oral outcomes due to tension free of the tongue.

**Discussion:** This paper presents a review about ankyloglossia, its symptoms and management, illustrated by two cases of severe ankyloglossia within in variation of age on intervention.

Keywords: Ankyloglossia; Tongue; Severe cases; Frenectomy; Blade surgery.

#### Introduction

Ankyloglossia or tongue-tie is a singularly congenital case qualified by a specific no elastic and dense frenum within lingual muscle. Also it's occurs when there is an union of the tongue's body to the mouth's floor, reducing her motion [1]. Its prevalence ranged of 0.02% to 12%. Lack of unified consensus may explain these rates changing on prevalence of ankyloglossia [2]. A higher incidence revealed in studies exploring neonates in comparison to those realized in other population was revealed by Reddy et al. 2014 [3]. The authors suggested the resolution of beginner variants of ankyloglossia during development justifying the age related difference of prevalence [4].

Ankyloglossia limits physiological tongue movement and may induce numerous functional, behavioral and speech difficulties [5,6]. In order to rectify these situations the treatment of ankyloglossia would requires frenal excision ensuring lingual

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motion [2]. The aim of this paper review illustrated with 2 clinical cases is to allow clarification of the basis, consequences and surgical management of severe tongue-tie.

# **Case presentation**

# Case I

A 14-years boy complains of lingual adhesion associated to speech trouble. In examination, the subject suffer from tongue-tie and his lingual mobility was limited (protrusion with a feeling of pulling, atypical swallowing, and inability of the tongue to reach retro palatal incisor region). The sign of severe ankyloglossia is clinically revealed by a deformity of the tongue and is classified as Class III using Kotlow's assessment (Figure 1). The challenge was dental crowding, malocclusion due to maxillary cleft and an atypical swallowing. No genetic implication was observed in the parents. `



**Figure 1:** Pre-operative view showing ankyloglossia class III: (a). with distorted tongue, (b). invagination at the tongue tip during the protrusion, (c). limited protrusion.

The management of this case is illustrated in Figure 2.



**Figure 2:** Operative view of frenectomy: (a). Incision using scalpel, (b). Completion of frenectomy.

Under local anesthesia, first an hemostat was clamped into the basis of the lingual frenum and followed by an incision at the upper side of the instrument. To close wound edges, superficial layer muscle fibres were dissected. Sutures were made to promote primary healing to reduce scar formation.

The post-operative period was asymptomatic without bleeding or pain (analgesic level 1 for 3 days). Sutures were removed after 7 days, lingual mobility was improved (Figure 3). Lingual rehabilitation and follow-up with the speech therapist was recommended for this patient.



Figure 3: Post-operative view: (a). Sutures, (b). follow up at 1 week.

# Case II

A 45-years male in apparently good general health, presented a short lingual frenum. The diagnosis show reduced lingual movements, and ankyloglossia Class III in Kotlow's classification (Figure 4). No genetic implication was declared by the patient and no intervention before was in taking.



**Figure 4:** Pre-operative view showing ankyloglossia class III: (a). lingual frenum pull, (b). limited protrusion of tongue.

Under local anesthesia, first an hemostat was clamped into the basis of the lingual frenum and followed by an incision at the upper side of the instrument. To reach easily closure superficial layer muscle fibres was dissected. Sutures were made to promote primary healing to reduce scar formation on the tongue's tip and buccal floor.

Just after the intervention, the patient was able to move more easily his tongue (Figure 5).



**Figure 5:** Operative view of frenectomy with immediate tongue liberation: a. sutures on the tongue, b. sutures on the buccal floor.

At the follow-up a better protrusion of the tongue and normal posture were possible without difficulty (Figure 6).



**Figure 6:** Post-operative view showing better protrusion of the tongue: (a). limited protrusion before surgery, (b). complete protrusion after surgery at 1 week.

## Discussion

At the beginning, a U-shaped split is formed on either sides of the tongue [7]. Before 8 years old, the body of the tongue gradually becomes free from the sides and the floor of the mouth [8,3]. A median fold of mucosal tissue is persistent near from the tip [3]. This procures lingual mobility, except at the region of attachment of lingual frenum [7]. Failure in apoptosis process of embryonic cells express fibrous and brief lingual frenulum linking tongue to its buccal floor [9].

Many morphological, anatomical and functional classifications have been cited in the literature [9]. The most used and simple is the Kotlow's classification [9]. According to its observations, we can define four types of ankyloglossia in relation to free tongue movement [3]. To achieve the diagnosis, a clinical-functional assessment has been suggested by Olivi et al. in 2012 [10].

In a review, Pompéia et al. in 2017 [11] concluded that clinicians approved negative effects of lingual frenum's anatomic and functional changes over craniofacial evolution and development. As described, numerous studies revealed link of those clinical outcomes and tongue mobility.

• Restriction of lingual frenulum and orthognathic development:

- The ankyloglossia can result in a reduction of maxillary development [12],
- A link in limited lingual movement with an elongation of the soft palate was demonstrated [13],
- The development of skeletal Class III malocclusion [8], and the predilection of mandibular prognathism were related to tongue-tie [3],

• Restriction of lingual frenulum and functions of the tongue:

The tongue tie is reveled as an obstructive sleep apnea a phenotype in children [14], it also maintain the atypical swallow [3], and may alter oral speech [15],

- Restriction of lingual frenulum and Oral hygiene:
- A short lingual frenulum may regularly limit the capacity of cleaning teeth. It seems to increase the prevalence of tooth decay [2],
  - Restriction of lingual frenulum and Periodontal health:

- Tongue tie can alter the gum and contribute to periodontal problems with apparition of gingival recession as described by Nathan in 2017 [16]. According to Veyssiere et al. in 2015 [9], this condition can be painful, may lead to poor oral hygiene, which in turn accelerate periodontal disease. In recent paper report of Bahadure et al. in 2019 [17], a rare and unique pattern of anky-loglossia where lingual frenulum was exceptionally attached and merged with lower labial frenum was described.

Managing tongue-tie is related to age, the location of the frenulum, the restriction degree, functional conditions, or social troubles [1,18]. In summary, current best practice should include frenectomy as a complete excision of the frenum with release of

lingual muscle fibers [6]. The aim is to separate the lingual body from its frontal join close to mucosal jaw [9]. This intervention gives maximum motion of the tongue's tip [16].The treatment of ankyloglossia can be realized via scalpel, electrocautery using multiple electrodes, or with laser surgery providing simultaneous cutting and cautery of soft tissue [16]. Even if laser offers advantages on surgery, some surgeons choose scalpel technique procuring more precise dissection. The variation on size of connective and fibrous compounds would determine the best technique of frenectomy [16].

According to literature, minor nerve or vascular vessels were objectified in middle of lingual frenum. In general, It's considered as a safe inferior plane as concluded by Hou et al. in 2012 [19]. The lingual topography and anatomy are at risk of multiple intraoperative or postoperative issues [6]. Among these complications, we can cite: hemorrhage, hematoma, paresthesia, infection. Also the re-establishment of frenal bond and scars may exerce a restriction in tongue movement [6]. Additionally, the lingual surgery on ventral area would induce Blandin-Nuhn gland mucocele, or blockage of Wharton's duct as described by Hungund et al. in 2013 [20] and Barot et al. in 2014 [18].

### Conclusion

Ankyloglossia can apply deleterious effect on daily life. Those worse outcomes can be explained by neglecting this congenital anomaly. It is suggested that early diagnosis and treatment of abnormal frenum are essential to optimal oral development. Different groups have to intervene for better tongue tie management.

## Declarations

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