**Ankyloglossia: Tongue Tie**

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**Background**

Ankyloglossia, from the Greek words “agklios” meaning curved and “glos-sa” meaning tongue, occurs in approximately 2-10% of live births\(^1\) with multiple studies reporting incidences around 4%\(^2\). The oral tongue is attached to the floor of mouth via a fold of tissue known as a frenulum which restricts the motion of the tongue. However, when the motion is overly restricted, the result is ankyloglossia. Ankyloglossia is also known as tongue tie and considered a minor congenital anomaly.

Normally, the tongue will extend and protrude out of the mouth without difficulty past the upper and lower alveolar ridges. In a patient with ankyloglossia, he or she can not extend the tongue past the alveolar ridges and/or is unable to lift the tongue to touch the palate. Tongue tie is not a new phenomenon. Horton details in a 1967 article, reports of midwives from ancient times tearing frenulums with dirty finger nails to release tongues\(^3\).

**Embryology**

During fetal development, the tongue and floor of mouth are fused. Through cellular degeneration and apoptosis, these structures separate. What tissue remains, is the lingual frenulum. When too much tissue remains, the result is ankyloglossia.

**Genetics**

The cause of ankyloglossia is generally unknown. However, there have been reports of possible familial inheritance in the literature\(^4,5\). One study reported the identification of a family with isolated ankyloglossia inherited in an autosomal dominant or recessive pattern in Iran\(^4\). Ankyloglossia has also been associated with a small number of rare disorders such as X linked cleft palate syndrome and van der Woude syndrome. Patients with X-linked cleft palate syndrome and ankyloglossia have been found to show mutations in the T-box transcription factor gene TBX22\(^6\).

**Epidemiology**

The incidence is generally reported in current literature to be around 4%\(^2\) but has been reported up to 10% in some studies\(^1\). There is a 2:1 male to female ratio of ankyloglossia according to multiple studies\(^2,7\) and can be diagnosed in neonates, children, and adults. There has been a wide range of reported incidence likely from the lack of uniform definition and differences in definitions used for diagnosis\(^1\).

**Diagnosis**

Ankyloglossia is usually diagnosed shortly after birth based on physical exam by a physician staffing the newborn nursery. A more detailed oral examination and subsequent diagnosis is usually prompted by difficulty feeding. The tongue is examined for extension, lateralization, ability to lift, cupping, and suck. Some cases will go undiagnosed until later in life and even adulthood.
Currently, there is no standardized definition regarding diagnosis of ankyloglossia. Proposed criteria for diagnosis have included length of lingual frenulum, amount of tongue movement, appearance of tongue tip being ‘heart shaped’, and palpation of fibrous tissue on physical exam (Figure 1 A to J).

Figure 1 A to J. Lingual frenulum examples

Similarly, multiple classification systems have been proposed but none universally accepted for use. Some classification systems rely on mainly on measurements of length of tongue tie. Others are more visual and can be based on where the frenulum attaches. One classification is the Kotlow classification which measures the distance of the insertion of the lingual frenulum to the tip of the tongue and is divided into four classes (Table 1).

Another classification system which is frequently used is based on how close to the tip of the tongue the leading edge of the frenulum is attached. There are four types which are listed in Table 2. Type 1 and 2 are considered classic tongue ties and obvious while type 3 and 4 are less common and more difficult to appreciate on physical exam. They often go undiagnosed.
**Table 1.** Four classes of Ankyloglossia ⁹.

<table>
<thead>
<tr>
<th>Class</th>
<th>Description</th>
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<tbody>
<tr>
<td>I</td>
<td>Mild ankyloglossia, 12-16mm</td>
</tr>
<tr>
<td>II</td>
<td>Moderate ankyloglossia, 8-11mm</td>
</tr>
<tr>
<td>III</td>
<td>Severe ankyloglossia, 3-7mm</td>
</tr>
<tr>
<td>IV</td>
<td>Complete ankyloglossia, &lt;3mm</td>
</tr>
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**Table 2:** Four types of Ankyloglossia.

<table>
<thead>
<tr>
<th>Type</th>
<th>Description</th>
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<tbody>
<tr>
<td>Type 1</td>
<td>Attachment of the frenulum to the tip of the tongue, usually in front of the alveolar ridge in the lower lip sulcus</td>
</tr>
<tr>
<td>Type 2</td>
<td>Attachment of frenulum two to four millimeters behind the tongue tip and attaches on or just behind the alveolar ridge</td>
</tr>
<tr>
<td>Type 3</td>
<td>Tongue tie is the attachment to the mid-tongue and the middle of the floor of mouth and is usually tighter and less elastic</td>
</tr>
<tr>
<td>Type 4</td>
<td>Essentially against the base of tongue, very thick, shiny, and inelastic</td>
</tr>
</tbody>
</table>

Some studies describe the classification of anterior and posterior tongue ties ¹¹. The anterior types are described as type 1 and 2 ankyloglossia as in the figure above. Posterior ankyloglossia is characterized as types 3 and 4.

Hong, et al made the diagnosis of posterior tongue tie when the lingual frenulum was not very prominent on inspection but was thought to be tight on manual palpation or was found to be abnormally prominent, short, thick or fibrous cord-like with the use of the grooved director ⁸.

**Sequelae**

The tongue is important in deglutination, mastication, and speech. One can imagine that if the tongue’s mobility is compromised from ankyloglossia, then pathology can result in those areas.

Concerns regarding early and late dysfunction as a result of tongue tie exist. Early dysfunction revolves around difficulty breastfeeding in newborn infants due to poor latch, ineffective milk transfer, excessive weight loss with failure to thrive, prolonged feeding times, and nipple pain and trauma to the mother. Late sequela can include difficulty with certain sound pronunciations, eating (unable to lick ice-cream cone), improper alignment of lower teeth at attachment of frenulum, difficulty playing wind instruments, and decreased oral hygiene from the inability to lick one’s lips or teeth from tongue tethering. Some of these problems listed have more of a social impact on the patient rather than medical but can be severe enough to warrant surgical correction.

**Tongue Tie and Breastfeeding**

In recent times, there has been much controversy surrounding the effects of ankyloglossia on breastfeeding especially given the renewed push for mothers to breastfeed neonates.
Breastfeeding is a neonatal reflex. Ankyloglossia may impede normal peristaltic movement of the tongue during breastfeeding. Marmet et al. noted a tight frenulum caused the tongue to move toward the floor of mouth quickly which could lead to an ineffective seal to the breast or induce the infant to grasp the breast more firmly with suckle; this can cause significant pain to the mother. However, one must keep in mind difficulty breastfeeding can be from numerous causes such as placental transfer of medication; lack of skin to skin contact between mother and baby; other anatomic anomalies such as with the palate, mandible, lip; and neuromuscular etiologies. It is important to evaluate for all possible causes leading to difficulty breastfeeding.

There are a number of studies evaluating tongue tie and breastfeeding. Some results show increased difficulties with breastfeeding neonates with ankyloglossia, especially within the first week of birth. Ricke, et al., who performed a case control study, found that mothers of babies with ankyloglossia were 3 times more likely to give up breast feeding and turn to the bottle. However, by one month, there was no significant difference between the number of breast feeding mothers with babies with ankyloglossia compared to those without ankyloglossia. In the same study, there were also a number of mothers with babies with ankyloglossia who reported absolutely no difficulty breast feeding. Furthermore, even when problems relating to ankyloglossia exist, the frenulum can stretch over time with no intervention, leading to improvement. A randomized control study by Emond et al. looked at the effectiveness of frenulectomy in treating ankyloglossia using LATCH (Latch, Audible swallowing, nipple Type, Comfort, and Hold) scores and found no breastfeeding improvements after frenulectomy. LATCH is a validated 10-point score with moderate predictive value for identifying mothers at risk for early weaning because of sore nipples.

Increased pain with breastfeeding has been associated with breastfeeding neonates with ankyloglossia. However, nipple pain is common with breast feeding. Buck et al. reported 79% of breastfeeding mothers surveyed reported nipple pain. This number dropped to 20% after 8 weeks of breast feeding (Buck).

A recent systematic review of the literature was conducted by Francis, et al with findings showing a small body of evidence suggesting that surgical correction of ankyloglossia may be associated with mother-reported improvements in breastfeeding, and potentially in nipple pain, but with small, short-term studies with inconsistent methodology, strength of the evidence is low to insufficient (Francis). Currently, there is no clear answer regarding the effects of tongue tie and breastfeeding and recommendations for treatment.

**Tonge Tie and Mobility and Speech**

Concerns arise regarding mobility and speech development in patients with ankyloglossia. The problems associated with mechanical limitation of the tongue can result in speech difficulties in pronunciation of consonants like t, d, n, and l, and make it difficult to roll an “r,” as these sounds require tongue tip elevation which can be inhibited in ankyloglossia to varying degrees. However, many patients with ankyloglossia have normal speech when evaluated by speech pathologists and clearly adapt to making these sounds well.
There are studies which evaluate tongue mobility and speech prior to and after surgical release. One such study by Messner, et al performed a prospective study with 30 children from 1 to 12 years of age with ankyloglossia undergoing frenulectomy. Parents of 83% of the patients believed their speech was adversely affected. Their results showed a significant improvement in tongue mobility and speech after frenuloplasty in children with ankloglossia who had articulation problems. According to Webb, however, there is insufficient data in the literature on any significant association between speech difficulties and ankyloglossia and therefore do not recommend tongue tie division in early infancy for this reason.

**Management**

Management for ankyloglossia includes conservative management of observation as well as surgical correction. Treatment depends on the treating physician, severity of symptoms from ankyloglossia, and parental and/or patient wishes depending on the age of patient.

Regarding treatment in neonates, many sources recommend observation rather than surgery unless the tongue tie is resulting in difficulty breast feeding along with significant weight loss in the neonate. It is thought that with time, the frenulum can stretch and/or the baby learns to compensate. No surgical intervention is recommended until at least 24 hours after birth and the mother and baby have worked with a lactation team, if available. Only after 24 hours and with evidence of minimal to no improvement in difficulty latching or pain should frenulectomy be considered. Buryk, et al recommends waiting until day of life 2-6 to establish the breastfeeding pattern. Surgical treatment is also recommended in children or adults with tongue tie if it results in difficulty with speech (ie pronunciation of certain consonants), but this should be evaluated by a speech therapist first whenever possible. Another indication is dentition where the bottom central incisors appear to be crooked. Oral hygiene is another concern. Eating, such as licking an ice cream cone, or social stigmas are softer indications.

**Surgical Treatment**

Surgical treatment has been around since ancient times. Literature from the 1800s describes midwives leaving one of their fingernails long in order to release the lingual frenulum after birth. The most popular and well known modern surgical treatment method for neonates involves performing a simple clipping of the lingual frenulum known as frenulectomy, also referred to as frenotomy or frenuloplasty by some.

The actual process of performing a frenulectomy on a newborn can be relatively quick and low risk compared to other surgical procedures with an approximate time of 3-5 minutes. Figure 2A shows the equipment needed for frenulectomy (gloves, oral sucrose, straight hemostat, iris scissors, 2x2 gauze pads and Figure 2B shows a grooved tongue retractor) and Figure 3 the frenulum identified. Prior to performing the procedure, it is imperative for the surgeon to review the risks and benefits of the procedure and what to expect after the procedure with the family. Risks include bleeding, infection, failure to improve breastfeeding, failure to prevent speech or dentition abnormalities, scarring with recurrence of symptoms,
need for repeat or further procedures, bleeding possibly requiring sutures, and injury to surrounding structures including the tongue, floor of mouth, and submandibular ducts. Once the risks and benefits of the procedure are reviewed with the parents and informed consent obtained, the neonate is usually transferred to a nearby procedure room by nursing staff if still in the hospital or performed in clinic.

First the baby is administered oral sucrose which may act as an analgesic for the neonate (Figure 4). Next, the surgeon uses his or her fingers to lift up the neonate’s tongue exposing the tongue tie or a groove director instrument (grooved tongue retractor Figure 2B) can also be used to retract the tongue and expose the lingual frenulum. Once adequate exposure and visualization is achieved, a small hemostat clamp is applied to the area of lingual frenulum to be clipped for approximately 30 seconds then removed (Figure 5). This helps to devascularize the area and decrease bleeding. Finally, small iris scissors are used to incise long the path of the clamp mark on the lingual frenulum (Figure 6 and 7) releasing the tongue tie while taking care not to injure the submandibular ducts nearby. There is usually mild bleeding which resolves quickly. The baby is returning to his or her parents and can immediately begin breast feeding.

In older children or adults, the procedure is usually performed under general anesthesia in the operating room using monopolar cautery. Some practitioners place sutures but these are not always necessary. However, there are reports of using CO_{2} and YAG lasers and other more complicated tongue tie releases such a Z-plasty or V-Y plasty techniques^{24-27}.
Complications

Complications from frenulectomy in ancient times included exsanguination from the lingual artery and infection leading to tongue swelling, possible asphyxiation and death. Luckily, these complications are generally unheard of in current times. The most frequent complication after simple frenulectomy is mild bleeding which usually self resolves or resolves with mild pressure; some cases may require a suture or two.23

There are rare case reports of patients developing infections or swelling after surgical treatment in areas such as the floor of the mouth and sublingual/submandibular regions, but again, this is very rare.28,29 Sirinoglu28 reported a case of submandibular swelling hours after performing a frenulectomy under general anesthesia on a 3 year old boy. This was thought to be from sutures placed which obliterated the Wharton ducts orifice. There was no airway compromise or difficulty eating. The patient was observed in the hospital with administration of intravenous antibiotics and anti-inflammatory medications, and the patient had an uneventful recovery.28
Conclusion

The problems associated with mechanical limitation of the tongue due to ankyloglossia are difficulties in breastfeeding among neonates, malocclusion, improper oral hygiene and speech difficulties.

In recent years, given renewed push for breastfeeding neonates, there has been renewed interest in effects of ankyloglossia and the need for treatment. Current literature is unclear regarding effects of ankyloglossia and breastfeeding. However, given there is evidence suggesting surgical correction can improve latch and potentially encourage mothers to breastfeed longer, and given the low risk of the procedure, frenulectomy should be considered in neonates with ankyloglossia. Many older patients with tongue tie are asymptomatic and require no treatment at all, but there are those with select sequela who may warrant consideration as well.

Ankyloglossia has been a known entity for a very long time. It comes in many different shapes, sizes, thicknesses, severities. Universal diagnostic criteria nor classification systems currently exists. Hopefully, a universal definition and classification system can be agreed upon in the near future which would assist in obtaining more objective data on the effects of ankyloglossia.

References