Randomized Controlled Trials Change Perspective on Ankyloglossia Management

The management of ankyloglossia in breastfed infants has changed thanks to new information from recent randomized controlled trials.

Ankyloglossia -and its remedy, the frenotomy- has been described in medical and historical documents throughout the centuries. Traditionally, frenotomies had been performed by primary care providers. They were performed because professionals believed that the presence of a lingual frenulum could result in speech and feeding problems.

Studies in the early twentieth century questioned the significance of the lingual frenulum, and therefore, the value of the frenotomy procedure. These studies suggested that the lingual frenulum did not typically cause speech problems. As a result, the practice of routinely performing frenotomies in newborns drastically declined. Additionally, infant formula was introduced around the same period in which these speech studies were published. This introduction resulted in a drastic decline in breastfeeding rates. By the mid-twentieth century, few babies were nursed. The impact of ankyloglossia on effective breastfeeding only resurfaced as breastfeeding rates increased toward the end of the 20th century. Within the past few years, three randomized control studies on the value of frenotomies in babies with ankyloglossia were published, all of which demonstrated a clinically significant reduction in infant feeding problems.

Definition

Ankyloglossia is a condition in which the function and movement of the tongue is compromised due to a structural restriction. This usually involves the presence of a lingual frenulum. However, ankyloglossia without the presence of a lingual frenulum occurs in very rare situations, where the tongue is directly fused to the floor of the mouth. It must be noted that the mere presence of a lingual frenulum only meets the definition ankyloglossia if tongue function and movement is compromised.

Classification

Ankyloglossia can be classified in several ways. In one useful approach, ankyloglossia is categorized according to the relationship of the most anterior portion of the frenulum tissue to the tip of the tongue (see Table 1). This classification helps an evaluator recognize that lingual frenula may be more obvious (such as when frenula are present most anteriorly) or less obvious (such as when frenula start at the very back portion of the tongue’s underside).
Ankyloglossia is also classified as anterior and posterior tongue tie. Anterior tongue tie is defined as when the anterior aspect of the frenulum starts anywhere from the tongue tip to just before the junction of where the tongue’s underside meets the floor of the mouth. This would include Types 1 through 3 of the previous classification. Posterior tongue tie is defined as when the frenulum is at the junction where the tongue’s underside meets the floor of the mouth. It correlates with Type 4 in the previous classification. Anterior tongue tie comprises 92% of cases. The male: female ratio is 3:1. Posterior tongue tie encompasses 8% of cases and is associated with a 3:1 female: male predominance.

**Incidence**

Studies show that the lingual frenulum is seen in 3-5% of people. 25-40% of nursing babies with a lingual frenulum will have a breastfeeding problem.

**Ankyloglossia and Associated Problems**

Ankyloglossia is occasionally associated with problems other than breastfeeding. It should be noted that a significant number of people with lingual frenula have no complaints and therefore do not technically meet the strict definition of ankyloglossia.

**Dental problems** may occasionally be associated with ankyloglossia. Frenula that are attached on the gum line may cause separation of the lower central incisors. Additionally, ankyloglossia can rarely result in poor manipulation of food boluses. The persistent presence of food material along the floor of the mouth poses a choking risk. Harboring food in the oral cavity also increases the risk of dental caries. In old age, the attachment of the frenulum on the lower gum line can interfere with the proper fitting of dentures.

**Social Problems** Occasional social problems have been reported, including impairment of licking food or trouble kissing.
**Speech Problems** It is widely accepted that ankyloglossia is not associated with speech delay, though its association with articulation problems is controversial. Though it is likely that some patients do experience articulation problems due their lingual frenula (particularly with sounds that require lifting of the tongue tip to the upper incisor-hard palate junction), larger studies and randomized control trials would need to be conducted in order to definitively answer this question.

**Breastfeeding Problems**

**Signs and Symptoms**

Nursing babies with ankyloglossia may have feeding problems. Their mothers may experience nipple pain. Problems with latch include difficulty with latch initiation, shallow latch and frequently slipping off the breast. Some infants make clicking noises as they partially lose suction with each suck. Milk retrieval inadequacy manifests as near-constant hunger with abnormally increased feeding frequency, poor weight gain and, in severe cases, failure to thrive. Maternal breast pain presents as sore nipples (see box below for definition of abnormal and physiologic nipple pain), nipple excoriation, areolar/ breast bruising and mastitis. Ballard, et al showed that breastfeeding problems worsen over time in mothers and their babies with unrepaired ankyloglossia. For example, poor milk retrieval may result in slow weight gain and eventually in failure to thrive.

<table>
<thead>
<tr>
<th>Classification of nipple pain</th>
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<tr>
<td><strong>Normal (physiologic) nipple pain:</strong></td>
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<tr>
<td>Discomfort goes away within 60 seconds of latch</td>
</tr>
<tr>
<td>Is completely gone within 2 weeks of age</td>
</tr>
<tr>
<td>No excoriations or bruising</td>
</tr>
<tr>
<td><strong>Abnormal (pathologic) nipple pain:</strong></td>
</tr>
<tr>
<td>Discomfort lasts more than 60 seconds after latching</td>
</tr>
<tr>
<td>Continues beyond 2 weeks of life</td>
</tr>
<tr>
<td>May or may not be associated with excoriations or bruising</td>
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**Research**

Hogan, et al published the first randomized controlled study. It compared immediate frenotomy to 48 hours of intensive feeding consultation in infants with ankyloglossia and who have feeding problems. Feeding improved in 27 of the 28 infants who received immediate frenotomy, whereas only 1 of 29 controls demonstrated feeding improvement following 48 hours of intensive feeding consultation. In the crossover phase of the study, as required by the study’s IRB, 28 of the 29 parents of the original controls consented to frenotomy. 27 of the 28 original controls, who had subsequently received frenotomy in the crossover phase, demonstrated feeding improvement. The authors concluded that the frenotomy procedure in infants with ankyloglossia and feeding problems improves feeding quality and is superior to intensive feeding consultation.
Buryk, et al published a single-blinded randomized control study in nursing infants with ankyloglossia. The study compared frenotomy to a sham procedure. Infants were selected using the Hazelbaker Tool for Lingual Frenulum Function (HTLFF), a tool that assesses the appearance and function of the tongues of infants with ankyloglossia. Those identified with ankyloglossia were randomly assigned to either a frenotomy (30) or a sham (28) procedure. An F test assured that an adequate number of subjects were entered into the study. Feeding problems and maternal nipple pain were assessed before and after the frenotomy or sham procedure. The mother was blinded to the procedure or sham until post-assessment was completed.

Infants who received frenotomy had a clinically significant improvement in feeding scores when compared to the sham procedure (p=0.029). Maternal Pain scores decreased in both groups, but the frenotomy group had a clinically significantly greater decrease in pain compared to the sham group (p<0.001). The study’s IRB required a crossover phase of controls. The parents of 27 of 28 controls subsequently consented to the frenotomy procedure. Feeding scores improved and maternal pain scores diminished in the original control group who had subsequently received the procedure in the crossover phase.

Berry, et al conducted a double-blinded randomized controlled study in nursing infants with ankyloglossia. Infants were randomly assigned to either a frenotomy (27) or a sham (30) procedure. A 2 group χ² test with 0.05 two-sided significance level assured that an adequate number of subjects were entered into the study. Feeding scores and maternal nipple pain scores were assessed before and after the frenotomy or sham procedure. The mother and observer were blinded to the procedure or sham. Thus, a different individual, who was not involved with the evaluation, performed either the frenotomy or sham procedure.

Feeding scores improved significantly only in the frenotomy group (p<0.02). However, there was no clinically significant difference in maternal pain scores. The authors suggest that the lack of difference in pain scores might be explained by an older mean age of three weeks in this study, as compared to other studies (the mean age in the Buryk study was 6 days). This might result in mothers with more significant nipple trauma compared to other studies. A more complicated pain assessment process may have been necessary to appropriately assess maternal discomfort.

Though further studies are needed to determine whether maternal pain is a primary or secondary sign associated with ankyloglossia, it is clear from these randomized controlled studies that infant feeding quality significantly improves in babies with ankyloglossia who have had a frenotomy.

A recent ultrasound study offers insight into the tongue mechanics of babies with ankyloglossia before and after frenotomy. Geddes et al evaluated infant latch, maternal pain, milk retrieval and feeding mechanics in 24 nursing infants with ankyloglossia. They found clinically significant differences in all of these parameters after frenotomy. Ultrasound demonstrated greater up and down movement of the posterior tongue, increased intraoral negative pressure and more milk flow post-frenotomy.

**Assessment**

**Identification of the Lingual Frenulum**

While frenula that originate at or near the tongue tip are easy to identify, those with attachments located more posteriorly along the tongue’s underside are more challenging to visualize. In such cases, use of the physical examination process outlined below helps.
The examiner sweeps a cotton swab from one side of the tongue’s underside to the other side in an arc-like motion. If the swab cannot move beyond the midline, it is highly likely that frenulum tissue is present. If a bump at the back of the tongue’s underside is felt with the cotton swab, it is possible that a posterior tongue tie might be present. This maneuver can also be performed by using the 5th finger instead of a cotton swab. However, infants seem to tolerate the cotton swab method better. The next step is to verify the presence of the frenulum by lifting the tongue. This can be achieved by either pushing the tongue back with the side of the cotton swab or by using a grooved director instrument (see below) to push the tongue back in order to expose the tongue’s underside.

**Oromotor Assessment**

An oromotor assessment helps practitioners select candidates who would benefit from the procedure from those who would not. Some use the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF). This tool is frequently used in research and is thought to have good validity. Amir et al found that one appearance component (describing attachment of the frenulum to the gum line) and three function components (tongue protrusion, tongue elevation and tongue lateralization) had good kappa values for inter-rater reliability. The HATLFF does require some practice. It takes some time to complete the tool.

Srinivasan et al suggested a brief assessment tool, known as the Frenotomy Decision Rule. The approach is to focus on babies with lingual frenula who are experiencing a feeding problem (infant latch or weight problems, maternal nipple pain or trauma). For those with a feeding problem, an oromotor assessment is performed. Tongue protrusion, tongue elevation and appropriate tongue cupping is assessed. Failure of at least one movement suggests that the procedure would likely help. This approach is easy to learn and takes little time. A modified approach to the frenotomy decision rule is suggested by this author. The oromotor movements are modified to assess the three tongue movements that Amir et al determined had good inter-rater reliability (tongue protrusion, tongue elevation and the transverse tongue reflex):

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**Modified Frenotomy Decision Rule**

Feeding problem identified
(latch problem, weight gain problems and/or maternal nipple pain)

Abnormal Oromotor Assessment
One or more of the following:
poor tongue elevation and/or tongue protrusion and/or abnormal transverse tongue reflex

Perform Frenotomy
**Tongue Elevation**
The infant should be able to lift the tongue at least halfway toward the roof of the mouth. This can be assessed by the following:
- Observation. The assessor makes a point of observing where the tongue’s position is if the baby happens to spontaneously open his or her mouth or cries.
- The assessor quickly touches the tongue tip, followed by rapidly brushing the finger on the upper lip. This is a particularly helpful maneuver in young hungry babies.
- The baby sucks on a gloved finger. The assessor then rapidly pulls the gloved finger out in an upward direction.

**Tongue Protrusion**
The assessor looks for tongue protrusion over at least the anterior rim of the lower alveolar ridge.
- Observation. The assessor makes a point of observing for spontaneous protrusion of the tongue.
- The baby sucks on a gloved finger. The assessor feels whether the lower alveolar ridge is directly tapping the finger. The lower lip is then pulled down while the baby is sucking on the finger in order to note whether the tongue moves past the gum line.
- The assessor touches the tongue tip and quickly and lightly sweeps the finger onto the lower lip.

**Transverse Tongue Reflex**
The transverse tongue reflex is a primitive reflex where the tongue follows a gloved finger from one side of the gum line to the other. This reflex is present until 6-7 months of age.
- The examiner simultaneously touches the lateral side of the tongue and the corresponding lower gum line with a gloved finger (both must be touched at the same time to elicit the reflex). The examiner then runs a gloved finger from one side of the lower gum line to the other and then back again. A normal tongue will follow the finger. If the tongue does not move along with the finger or if it only moves on one side, the tongue movement is considered abnormal. A tongue that “catches” or tilts is also considered abnormal.
- It is helpful to practice eliciting this reflex on normal babies. This will help practitioners get a sense of how easily the reflex is elicited.

**FRENOTOMY**

**The Role of Primary Care Physicians**
Primary Care Physicians have performed frenotomies for centuries. From a technical point of view, the frenotomy procedure is simpler and more straight-forward than many other procedures that primary care physicians perform in their offices. The Academy of Breastfeeding medicine states that primary care physicians may consider performing the procedure in babies who are under 4 months of age, with anterior tongue tie and who have a translucent frenulum. The recommendation of 4 months is somewhat arbitrary but is primarily based on the ease of performing the procedure. Anterior tongue tie frenotomies are easier to perform when compared to posterior tongue ties. The frenula of patients with posterior tongue ties can occasionally be fibrous, whereas the frenula of patients with anterior tongue patients are thin and soft. Patients with posterior tongue tie should be referred to otolaryngologists, dentists, oral surgeons or certain breastfeeding medicine specialists.
Frenotomies can be performed in hospital nurseries or in the office. Infants as young as one to two days old are eligible candidates if it is determined that significant feeding issues are attributed to ankyloglossia. Ballard et al demonstrated that earlier correction through frenotomy is associated with fewer complicated feeding problems.

Candidates should have a white, translucent appearance upon compression with a finger. In contrast, a reddish appearance or pink strands within the frenulum likely represent entrapped genioglossis muscle from the floor of the mouth. This is an extremely rare occurrence. Patients with a reddish non-blanching frenulum or with pink strands within the frenulum should be referred to otolaryngologists, dentists or oral surgeons.

Definitions
Frenotomy is defined as the division of the frenulum without repair. It is synonymous with frenulotomy. Frenuloplasty is defined as division of the frenulum with repair (usually z-plasty). While frenotomies are typically performed on babies, the frenuloplasty is the procedure of choice in children and adults. The remainder of this article will focus only on the frenotomy procedure.

Complications
Few complications from frenotomy are reported in the modern literature. Theoretical potential exists for infection and bleeding. It is postulated that infection rates are extremely low because sterile instruments are used and because a host of anti-infective products are in human milk. The frenulum of infants has a low blood supply. Typically, only scant bleeding is noted following incision. Other potential complications such as glossoptosis have only been described in those undergoing a frenuloplasty (not frenotomy) under anesthesia.

The Procedure
Several techniques may be used to perform a frenotomy. The most common approaches include the instrument–grooved director, instrument–hemostat and finger-stabilizing techniques.

Regardless of chosen technique, an assistant should ideally be present to stabilize the baby, including gently holding the jaw still.

Grooved Director Instrument Technique
The physician uses an instrument known as a grooved director (also referred to as a grooved retractor or Lorenz retractor). This instrument pulls the tongue back, exposing the lingual frenulum.

The physician pulls the tongue back using the grooved director in the nondominant hand and cuts the frenulum with straight iris scissors. Some prefer to make two small cuts down to the floor of the mouth while others make one single cut. Others will cut almost to the mouth’s floor and then use a finger to slightly finger dissect to the floor of the mouth.

The advantage of using a grooved director is that it allows for firm control of the tongue and shields structures located in the back of the mouth, such as the Wharton’s ducts.
These instruments are often found in circumcision kits, though they are almost never used for circumcision. They can be ordered separately and individually packaged.

**Hemostat Instrument Technique**
The physician uses a hemostat to clamp the lingual frenulum along the line of anticipated incision. The clamp is held in place for about a minute and then released. A track from the clamp is then visualized. The physician then uses straight iris scissors, cutting along the track line.

The advantage of this technique is that the hemostat crushes blood vessels, resulting in minuscule bleeding. The disadvantage is that viscous lidocaine must be used prior to the procedure. The numbing of the oral cavity means that it will not be possible to adequately assess oromotor movement and feeding for about 45 minutes following the procedure.

**Finger-Stabilizing technique**
The physician uses one or two fingers of the nondominant hand to stabilize the tongue, cutting the frenulum with iris scissors using the dominant hand.

There are two ways to use the finger stabilizing technique. Some will use the second and third fingers of the nondominant hand, oriented in an upside-down V position, to pull the tongue back. Others prefer to use the index finger of the nondominant hand to push the tongue up and slightly to the side.

**Pain Control**
It is thought that frenulum tissue in young infants has minimal innervation. Traditionally, topical anesthetic is not used in anterior tongue tie frenotomies for this reason and because the procedure is done very rapidly. However, viscous lidocaine is used before performing posterior tongue tie frenotomies because they may take more time to perform and are slightly more complicated. Additionally, those who use the instrument-hemostat technique routinely apply viscous lidocaine because the hemostat is clamped for about a minute and because the tissue is crushed in the process. If viscous lidocaine is applied to the frenulum and surrounding tissue, assessment of nursing may not be possible for up to 45 minutes. The infant may not be able to fully control tongue movements while latched.
Post-Procedural Care
The incision is compressed with gauze until the bleeding stops. Usually bleeding is scant and brief. Oromotor movements are reassessed. The baby is then offered the breast. 90% of the time mothers notice a difference in feeding or discomfort. If a mother has a nipple erosion, it may take up to a week to determine if the procedure helped. The parents should know that a white eschar will develop at the wound site and will disappear within 3 weeks. Parents follow up in 3-4 days.

A mother with significant nipple and areola erosions will need to heal. Advise her that maternal intake of acetaminophen and ibuprofen will not harm her baby. Though some sources advise mothers with intensive pain to pump breasts 4-5 times a day while in the process of healing, this may lead to milk supply issues. Scabs often open as they get sucked off by the baby or the pump. Consider advising her to use an ultrathin nipple shield for 1-2 weeks instead. This often takes the edge off of the pain and allows the erosion to heal because the ultrathin nipple shield creates a barrier between the scab and the baby’s mouth. If the baby develops a preference for nursing with the shield, certain measures can be taken. Once the scab is completely gone and the skin has totally healed, the mother can start the feeding with the shield and make it fall off halfway through the feeding. Many babies are relaxed by this point and will then latch directly on the breast by that point in the feeding. After a week of success with this, the mother should see if the baby will latch without the shield.

Though most babies thrive when their mothers use an ultrathin nipple shield, it is probably prudent to keep track of the baby’s weight on a weekly basis.

A few older infants may have developed poor sucking habits prior to correction of the ankyloglossia, that need undoing (though most infants latch very well after the frenotomy is performed). Suck training entails holding the baby by the mother’s chest in position to feed. The mother allows the baby to suck on her 5th finger for 30-60 seconds. Upon removal of the finger the mother immediately presents the infant with her nipple areolar complex with the intention that the baby’s sucking and rooting reflexes will then help him or her latch. If suck training is not successful, the mom and baby may have to temporarily use an ultrathin nipple shield, following the procedure outlined above.

Malpractice Coverage
In general, most insurance carriers consider frenotomy to be within the scope of practice of primary care physicians. It is obviously important to verify this. Physicians may need to clarify with the carrier that the intention is to perform frenotomies (incision without repair) rather than frenuloplasties (incision with repair).
Coding
The code for frenotomy is 41010. The ICD 9 code for ankyloglossia currently is 750.0. The ICD 10 code for ankyloglossia will be Q38.1. Related breastfeeding codes are listed in the table below.

<table>
<thead>
<tr>
<th>ICD 10 (in 2014)</th>
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<tr>
<td><strong>Q38.1</strong> Tongue-tie, Ankyloglossia (was 750.0)</td>
</tr>
<tr>
<td>R63.3 Infant feeding problems (was 783.3)</td>
</tr>
<tr>
<td>Newborn feeding problems:</td>
</tr>
<tr>
<td>P92.2 Slow feeding of the newborn</td>
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<tr>
<td>P92.3 Underfeeding of newborn</td>
</tr>
<tr>
<td>P92.5 Neonatal difficulty feeding at the breast</td>
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<tr>
<td>P92.8 Other feeding problems of the newborn</td>
</tr>
<tr>
<td>P92.9 Feeding problems of newborn unspecified</td>
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[Note Incision of the lingual frenulum stays as 41010]

Summary
Recent randomized controlled studies demonstrate that feeding problems associated with ankyloglossia are improved following the frenotomy procedure. This procedure is within the primary care physician’s scope of practice.

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