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Otolaryngology (D Sidell and EM Arjmand, Section Editors)

Tongue Tie in Infancy

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Opinion statement

Ankyloglossia, or "tongue tie," classically involves a short or thickened lingual frenulum that may prohibit tongue protrusion. However, the diagnosis, evaluation, clinical significance, and management of ankyloglossia are widely variable and controversial. Despite attempts to create standardized diagnostic criteria for ankyloglossia, there has yet to be a universally accepted system. Management of ankyloglossia often includes a multidisciplinary approach including lactation consultants, speech language pathologists, pediatricians, and otolaryngologists. Observation or conservative management for asymptomatic infants or infants with minimal or well-compensated symptoms is a reasonable option, whereas surgical intervention may be warranted for infants and children with ankyloglossia that has significant impact on breastfeeding or speech. Frenotomy (also known as frenulotomy) is a relatively simple procedure that can be performed at the bedside or office setting in very young infants, precluding the need for general anesthesia. Frenuloplasty is usually performed on older children and should be performed in the operating room with general anesthesia. Overall, there is insufficient evidence to definitively associate ankyloglossia with breastfeeding or speech deficits. Surgical intervention for ankyloglossia should be recommended with caution and performed only on infants or children with clear findings of ankyloglossia on physical exam and a documented history of breastfeeding or speech difficulties; the timing and method of treatment should be tailored to the individual infant or child. Frenotomy or frenuloplasty should only be performed by providers with adequate training and experience in order to minimize complications.

Introduction

Ankyloglossia, commonly known as "tongue tie," etymologically originates from the Greek "agkilos" (curved) and "glossa" (tongue) and refers to congenital oral anomalies with varying degrees of restricted tongue

mobility. Characteristically, ankyloglossia involves an abnormally short or thick lingual frenulum or a highly attached genioglossus muscle insertion that tethers the tip of the tongue and prevents tongue protrusion. Since the initiation of the World Health Organization's movement towards reliance on breastfeeding during an infant's first year of life, there has been significant interest in infants affected by ankyloglossia [1]. Nonetheless, the clinical significance, diagnosis, classification, and treatment of ankyloglossia all remain controversial [2–5]. Breastfeeding challenges as a result of ankyloglossia include difficulty with latching, maternal nipple pain, and insufficient feeding, which may put the infant as risk for early infant weaning or failure to thrive in more extreme cases [6]. Later in life, the restrictions on tongue mobility caused by ankyloglossia may include cuts from teeth on the frenulum, difficulties with licking lips, and more socially debilitating problems with speech articulation; however, definitive studies on these limitations are lacking [6–8].

Messner et al. (2000) demonstrated significant variation in opinion regarding the clinical significance of ankyloglossia and indications for treatment [9]. Sixty-nine percent of surveyed lactation consultants believed that ankyloglossia was frequently associated with feeding problems, while the same was reported by only 6 % of surveyed otolaryngologists and 1 % of pediatricians. Regarding speech difficulties, 60 % of otolaryngologists believed that ankyloglossia was at least sometimes associated with speech difficulties, compared to only 23 % of pediatricians and 50 % of speech language pathologists. Moreover, the need for surgical intervention was controversial, with surgery being recommended for feeding, speech, and social/mechanical reasons by 53, 74, and 69 % of otolaryngologists, respectively, compared to only 21, 29, and 19 % of pediatricians.

Epidemiology

The reported incidence of ankyloglossia among infants ranges from 0.1 to 10.7 %, with studies investigating oral mucosa abnormalities finding a lower prevalence of tongue tie (0.1–4.4 %) than studies looking at ankyloglossia alone (4.2–10.7 %) [6, 9–12]. A definitive incidence in the literature has been elusive, likely secondary to the absence of standardized diagnostic criteria. In regards to gender, there appears to be a slight male predilection for ankyloglossia over females, though some studies have also found equal gender ratios. Race and ethnicity have not been found to be predisposing factors [6].

Etiology

The etiology of ankyloglossia is generally unknown. The vast majority of infants with ankyloglossia are healthy infants without evidence of other congenital anomalies [12, 13]. Nonetheless, a few case series have found an association between ankyloglossia and rare congenital syndromes such as X-linked cleft palate syndrome [14], Kindler syndrome [15], van der Woude syndrome [16], and Opitz syndrome [17]. A genetic basis of ankyloglossia has also been suggested, as several studies investigating factors associated with ankyloglossia have found a positive family history among 10–53 % of patients [7, 8, 13]. Familial pedigree studies have also found evidence to support genetic transmission of ankyloglossia, though the inheritance pattern has yet to be clearly established [12, 13, 18]. Of particular focus in the literature is the T-box transcription factor gene TBX22, which has also been implicated in X-linked cleft palate [19]. The TBX22 gene in humans is located on the long arm of the X chromosome and expressed through eight coding exons related to the development of the posterior palatal shelves and the caudal tongue [20, 21]. Mutations of the TBX22 gene have been found in patients with cleft palate and ankyloglossia [19, 20, 22–24]. Maternal cocaine use during pregnancy has also been associated with ankyloglossia. A case–control series by Harris et al. (1992) found that partial ankyloglossia was 3.2 times higher among children exposed to cocaine in utero [25].

Diagnosis

The diagnosis of ankyloglossia is particularly challenging. Although several classification systems have been proposed, none offer correlation with functional impairments, and there is currently no universally accepted system [2–5]. Similarly, the functional limitations are difficult to assess as neither the maternal nipple nor the infant's tongue are visible during breastfeeding. Careful assessment by manual examination of the undersurface of the tongue and the attachments of the frenulum is necessary to reveal limitations in mobility. Anterior ankyloglossia is more readily apparent to the eye due to the prominence of a shortened frenulum and tethering of the tip of the tongue. Anterior ankyloglossia is characterized by insertion at the tip of the tongue (type

I) or slightly behind the tip (type II). Posterior ankyloglossia is characterized by a thickened frenulum (Type III) or a submucosal frenulum presenting as a flat, broad mound absent of any typical protruding frenular tissue, and restricting base of the tongue mobility (type IV) [4, 26, 27]. With more subtle physical findings and functional impairment, posterior ankyloglossia poses an even further diagnostic challenge, and due to a paucity of available literature, its incidence and relevance remains controversial [28, 29•, 30].

Treatment options

Frenotomy

Frenotomy, or frenulotomy, is a quick procedure that is performed relatively easily to correct both anterior and posterior ankyloglossia. For young infants, the procedure is often performed at the bedside or in the office, often by otolaryngologists. Generally, the tongue is superiorly retracted to expose the lingual frenulum which is then sharply incised, usually with sterile iris scissors. Care should be taken during the procedure to avoid the sublingual and submandibular gland openings along the floor of mouth by cutting the frenulum close to the ventral surface of the tongue. The incision is not sutured. There is usually very little blood loss following the procedure; any minor amount of bleeding can be controlled with localized pressure. The infant is allowed to feed immediately following the procedure.

There have been five randomized controlled trials [31–33, 34••, 35] and seven systematic reviews [36••, 37••, 38••, 39••, 40••, 41, 42] investigating the effects of frenotomy on breastfeeding. However, experts acknowledge that these existing studies alone are largely insufficient to support a clear relationship between frenotomy and improvements in breastfeeding, with large variations in diagnosis of ankyloglossia and methods of frenotomy procedure, low-quality of evidence from case–control studies and case series, and a notable lack of data on nonsurgical interventions for ankyloglossia. Nonetheless, randomized controlled trials have found that frenotomy does improve breastfeeding immediately or within 5 days of the procedure when compared to sham surgery or observation [32, 33, 34••]. Among 55 patients randomized to frenotomy compared to 52 patients without intervention, Emond et al. (2014) found improvements in maternal self-efficacy measures of breastfeeding but found no improvement of frenotomy on the *L*atch, *A*udible swallowing, nipple *T*ype, *C*omfort, *H*old (LATCH) Scale of breastfeeding [34••].

The impact of frenotomy on reduced maternal nipple pain during breastfeeding has been inconsistent. Buryk et al. (2011) found in a randomized controlled trial that measures of maternal nipple pain decreased significantly immediately following the procedure compared to infants treated with sham procedure, but these improvements in nipple pain were not demonstrated at 2-week or 2-month follow-up [33]. Similarly, Dollberg et al. (2006) also found immediate relief in nipple pain following frenotomy [35]. However, Emond et al. (2014) and Berry et al. (2012) found no differences in maternal nipple pain scores following frenotomy [32, 34••].

A systematic review by Chinnadurai et al. (2015) concluded that there is insufficient evidence to assess the efficacy of frenotomy on nonbreastfeeding outcomes such as speech or social outcomes. Chinnadurai et al. (2015) found that frenotomy did tend to improve bottle feeding and tongue mobility in

social situations such as eating ice cream, while there were limited cohort studies suggesting an improvement in speech articulation and intelligibility following frenotomy [43••]. A systematic review by Webb et al. (2013) also found insufficient evidence to support frenotomy for the treatment of speech articulation [40••].

The timing of frenotomy in the available literature has been variable, with ages at the time of procedure ranging from 1 to 18 days for those experiencing breastfeeding difficulties in the neonatal period [37••]. Frenotomy is typically performed at the bedside or in the outpatient clinic setting. General anesthesia is usually not necessary for those under 3 months of age [44]. Utilization of frenotomy in an outpatient clinic setting without general anesthesia has been found to be safe and cost-effective, with high parent satisfaction scores [45–47]. In a survey study of patients and/or guardians by Klockars and Pitkaranta (2009), frenotomy with no or local anesthesia was found to be safe and cost-effective [18]. Some case series report the use of local anesthesia or sucrose during the procedure [37••, 47, 48•]; however, many do not mention this technique, and even advocate against the use of local anesthetic [10, 33, 35, 49]. Ovental et al. (2014) found that topical benzocaine was not beneficial in decreasing crying time, which was used as a measure of infant pain [50•].

Frenotomy has been shown to be largely safe procedure with the majority experiencing no complications [38••, 47]. The most frequently reported complication is minor bleeding which is usually addressed with local pressure for hemostasis. Injury to the submandibular ducts, surgical site infection, and sublingual hematoma are rarely reported [37••]. Ulceration at the surgical site and delayed wound healing have also been reported [47]. Rates of reoperation range from 0.1 to 27 % among available case series, though many of these case series include both infants and children, with higher rates of reoperation in children [38••]. In order to minimize the risk of complications, frenotomy should only be performed by experienced and trained practitioners. However, in a survey of 425 North American physicians, 22 % reported performing frenotomy, while only 10 % were formally trained to perform the procedure [51]. Recurrence and necessity for procedural revision, secondary to inadequate release of the lingual frenulum or development of scar tissue, may occur more frequently in cases of posterior ankyloglossia than anterior cases, with revision rates of 21.1 versus 3.7 % respectively reported by Hong et al. (2010) [28].

A handful of studies have investigated the utility and safety of CO_2, argon, and Nd:YAG laser technique in frenotomy, though the use has been primarily demonstrated among older children and adults [52–55]. At this time, experience with laser frenotomy in infants is exceedingly limited. Those that advocate laser frenotomy report that the use of laser allows for improved surgical precision with minimal bleeding, decreased wound contraction and scarring, no need for suturing, and improved postoperative pain [56–58]. In these studies, laser frenotomy has been successfully performed as an outpatient and/or in the clinic setting. Although the use of laser appears to have some advantages, there are notable associated risks. Protective eyewear is necessary for the surgeon and surgical assistants, who all need to be properly trained in laser safety procedures. The patient's skin, teeth, oral tissues, and airway are at risk of accidental laser injury. Thus, while laser frenotomy does appear to be a reasonable alternative to conventional surgical methods, it has yet to become widely accepted among otolaryngologists. This may be attributable to the relative ease of performing

conventional surgical frenotomy among infants without risk of the potential complications of laser use.

Frenuloplasty

Frenuloplasty is a generally performed to treat ankyloglossia in older infants between 1 and 2 years of age, as well as older children and adults. For infants and young children undergoing frenuloplasty, treatment under general anesthesia would be required. General anesthesia can be achieved with either intravenous sedation and/or intermittent mask ventilation. Frenuloplasty is preferred for these children over frenotomy in order to decrease the risk of scarring and need for reoperation. Nonetheless, frenuloplasty may still result in scar contracture and persistent tongue tethering.

A traditional frenuloplasty is performed as a horizontal-to-vertical plasty. In this technique, the tongue is retracted superiorly to expose the lingual frenulum. The lingual frenulum is then sharply incised along the ventral surface of the tongue. As with frenotomy, care is taken to avoid injury to the submandibular ducts. For some patients, the genioglossus muscle may have to be incised for adequate release of the ankyloglossia. The defect along the ventral surface of the tongue is then closed primarily in a single layer using absorbable sutures.

An alternative method of frenuloplasty is a Z-plasty technique which serves to release the lingual frenulum and increase the length of the ventral tongue scar [49]. In a classic Z-plasty, opposing 60° triangles are created and rotated to lengthen the scar. The lingual mucosa is then closed with absorbable sutures. Comparing frenotomy to Z-plasty frenuloplasty among infants and children ranging from 5 days to 8 years of age, Yousefi et al. (2015) found that frenuloplasty had significantly greater improvements in speech articulation, reduced maternal breast pain, increased lingual mobility, and parent satisfaction. Heller et al. (2005) advocated using a 4-flap Z-frenuloplasty and reported significant improvements in frenulum length, tongue protrusion, and speech articulation compared to traditional horizontal-to-vertical frenuloplasty among children 3–9 years of age [59]. Some have also proposed that in ankyloglossia, the foreshortened lingual frenulum coincides with contracture of the genioglossus muscle. Thus, Choi et al. (2011) advocate the combination of Z-plasty with genioglossus myotomy [60]. After incising the ventral tongue mucosa, Choi et al. (2011) describe using electrocautery to horizontally incise and release the tightened genioglossus muscle. These investigators found no difficulties with tongue movement or articulation following genioglossus myotomy and reported improved speech outcomes.

Supplementing with a breast pump

Within current literature, there is a paucity of data available regarding nonsurgical methods of managing ankyloglossia. In a retrospective review of 287 pediatric surgery outpatients with ankyloglossia, Wright (1995) reported that 16 patients did not require intervention, 66 deferred the decision for management at the initial consultation and ultimately did not undergo surgery, and 101 were referred to speech pathology, of which 49 did not require subsequent operation, for a total of 46 % of patients that did not require surgical intervention. Riskin et al. (2014) surveyed Israeli mothers with infants affected by ankyloglossia and reported that these mothers were more likely to use a breast

pump to supplement ineffective breastfeeding [61•]. Infants with ankyloglossia can be fed expressed breast milk from a bottle until they are able to transition to breastfeeding, usually around 6–9 weeks of age.

Finger feeding

Finger feeding using feeder-controlled methods (Hazelbaker FingerFeeder or feeding tube in a bottle) can also be utilized [62]. The feeder's finger and the feeding tube are held inside the infant's mouth across the lips. This method works via operant conditioning to train the infant in appropriate feeding techniques. Feeding starts when the tongue tip protrudes over the alveolar ridge and the posterior tongue drops slightly. Feeding with the fingerfeeder stops when the tongue retracts or when the posterior tongue pushes against the feeder's finger.

Nipple shield

Nipple shields may also be used during breastfeeding, particularly wider nipple shields, though for infants with severe tongue retraction from ankyloglossia, the use of a nipple shield may exacerbate nipple pain by stimulating the infant's bite reflex.

Lactation consultation

A multidisciplinary approach may be beneficial in the management of ankyloglossia. Involvement of lactation consultants should be undertaken to help mothers find optimal latch positioning while breastfeeding. Riskin et al. (2014) found that a lactation consultant helped mothers of infants with ankyloglossia with breastfeeding problems [61•]. Infants with ankyloglossia are unable to bring their tongues down into the mouth to allow grasp of the nipple. A tight, diagonal grasp of the infant against the mother's body or a semi-reclined position of the mother may allow the infant to gape widely and form a deep latch to aid in expressing milk. Modifications to bring the infant's chin and tongue closer to the breast may also be helpful. Denting the breast at or just below the areolar margin with a finger forms a hollow for the infant's chin, which then allows for closer latch of the infant's lower lip and tongue onto the nipple [62].

Tongue exercises

Tongue exercises have been proposed for infants that do not undergo surgical intervention and for postoperative patients to prevent scarring. While oral exercises alone are unlikely to cure limitations of severe ankyloglossia, participation in tongue exercises specifically designed by a lactation consultant or occupational therapist may help to train the infant in tongue extension and protrusion as well as limit maladaptive compensatory tongue movements. Exercises primarily focus on tongue elevation and protrusion [62, 63]. Caregivers are instructed to insert their finger underneath the infant's tongue and perform sweeping motions as well as direct pressure upwards and downwards to facilitate elongation of the frenulum [62]. Massage of the undersurface of the tongue has also been posited to help loosen tissue and promote increased tongue mobility. Another commonly used strategy is the gradual removal of the nipple or pacifier from an infant's mouth while he or she is actively sucking to encourage protrusion and elevation of the tongue when trying to maintain contact with the object.

In the postoperative period following frenotomy or frenuloplasty, tongue exercises are also often utilized to help prevent scarring and recurrence of ankyloglossia. Recommended exercises include protruding the tongue in and out of the mouth repetitively, touching the tongue to the posterior aspect of the upper teeth while the mouth is open widely, moving the tongue side-to-side, and sweeping food back and forth in the mouth with the tongue [7].

Conclusion and pediatric considerations

There is currently insufficient evidence to clearly associate ankyloglossia with difficulties in breastfeeding or speech due to large variations in diagnosis of ankyloglossia and outcomes measures. However, there are a number of studies that suggest surgical intervention through frenotomy or frenuloplasty is beneficial in improving breastfeeding efficacy. It is unclear at present if treating ankyloglossia improves latch and reduces maternal nipple pain during breastfeeding, as very little information exists on alternative, nonsurgical methods of managing ankyloglossia. Surgical intervention should be recommended with caution only in infants for whom ankyloglossia is clearly evident on physical examination and who have documented difficulties with breastfeeding. Procedures to treat ankyloglossia should only be performed by trained, experienced providers.

Compliance with Ethical Standards

Conflict of Interest
Sophie Shay, Rachel Mandelbaum, and Nina Shapiro declare that they have no conflict of interest.

Human and Animal Rights and Informed Consent
This article does not contain any studies with human or animal subjects performed by any of the authors.

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The authors concluded that there is insufficient evidence to assess the efficacy of frenotomy on nonbreastfeeding outcomes such as speech or social outcomes. They also found that frenotomy improved bottle feeding and tongue mobility in social situations such as eating ice cream, while there were limited cohort studies suggesting an improvement in speech articulation and intelligibility following frenotomy.

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