# Ankyloglossia and Other Oral Ties

Jonathan Walsh, MD[a], Margo McKenna Benoit, MD[b,*]

**KEYWORDS**

• Ankyloglossia • Tongue-tie • Lip-tie • Frenulectomy • Frenotomy

**KEY POINTS**

- Ankyloglossia, or tongue-tie, has become a topic of great interest and some controversy over the past 20 to 30 years, as rates of breastfeeding initiation have increased.
- Tongue-tie can result in various degrees of difficulty with breastfeeding, oral hygiene, speech, and dentition.
- Diagnosis must include a functional assessment of tongue mobility, in addition to the physical appearance of the frenulum.
- Procedures to address ankyloglossia and other oral ties are commonly accepted as safe; however, serious complications such as severe bleeding, infection, and worsening glossoptosis have been reported.
- There is little evidence to date supporting surgical intervention for maxillary, mandibular, or other oral ties.

## INTRODUCTION

The lingual frenulum is formed during the fourth week of gestation as the 2 lateral lingual swellings move medially to fuse with the tuberculum impar, forming the anterior two-thirds of the tongue. The tongue then separates from the floor of mouth to form the lingual sulcus. Failure of release results in varying degrees of ankyloglossia, or tongue-tie, in which a fibrous band in the midline tethers the tongue to the alveolar ridge or floor of mouth. Ankyloglossia can be asymptomatic or may have a wide array of consequences, including difficulty with breastfeeding, oral hygiene, dental development, speech, and other social factors. Treatment often involves division of the band to provide better tongue mobility. Although this is generally accepted to be a minor and safe procedure, there is considerable debate about when and how to intervene. In addition, several case reports have documented serious and potentially

Disclosure Statement: The authors have nothing to disclose.
[a] Department of Otolaryngology–Head and Neck Surgery, Johns Hopkins University, 601 North Caroline Street, 6th Floor, Baltimore, MD 21287, USA; [b] Department of Otolaryngology, University of Rochester Medical Center, 601 Elmwood Avenue, Box 629, Rochester, NY 14642, USA
* Corresponding author.
*E-mail address:* Margo_Benoit@urmc.rochester.edu

Otolaryngol Clin N Am 52 (2019) 795–811
https://doi.org/10.1016/j.otc.2019.06.008
oto.theclinics.com
0030-6665/19/

life-threatening complications associated with frenotomy. In addition to anterior and posterior lingual ties, there is growing interest in clinical practice and in the literature on how best to evaluate and manage upper lip-tie (ULT) and other oral ties. Within the multidisciplinary community of providers interested in ankyloglossia, there exists a need for a solid understanding of the available literature and best practice recommendations to make discerning diagnostic and treatment decisions for patients. This article presents an evidence-based approach to ankyloglossia and other oral ties.

## HISTORY AND EPIDEMIOLOGY

Ankyloglossia, commonly known as tongue-tie, is an anatomic variant of tongue anatomy that has been recognized for centuries. Some of the first references to a disorder of the tongue being tethered to the floor of mouth appear in days of Aristotle in the third century BC, and the operative technique of dividing a tongue-tie was first described in the seventh century AD. In the middle ages, debate first arose between midwives, who used a long fingernail to perform the procedure, and surgeons, who were allowed to use instruments. In medieval times up and through the early 1900s, tongue-ties were released routinely. In fact, the instruments necessary to clip a tongue-tie appeared in circumcision trays because both procedures were commonly performed before a newborn was sent home from the hospital.[1,2] Around 1950, the culture in the United States changed with the introduction of baby formula and rates of breastfeeding fell dramatically. By the 1960s and 1970s, ankyloglossia was considered to be an outdated topic, and many pediatricians denied that tongue-tie existed or that it caused a problem with feeding in newborns.[3] The natural childbirth movement in the 1970s brought renewed interest in breastfeeding as a first choice for infants, and with this change came increasing recognition of tongue-tie as a potential road block to successful breastfeeding. A few case reports and observational studies appeared, proposing a link between ankyloglossia and breastfeeding difficulty. In the past 20 years, ankyloglossia has become a controversial topic in medicine, with many strong opinions held by a diverse group of providers, including pediatricians, neonatologists, feeding and speech therapists, lactation consultants, dentists, and otolaryngologists.[4]

Walsh and colleagues,[5] in a study of the KIDS database, reported an increase in the diagnosis of tongue-tie by 834% from 1997 to 2012 and a similar increase in the number of frenotomy procedures performed over that same time period. Similar epidemiologic studies in Australia and Canada have shown a marked increase in ankyloglossia diagnosis and frenotomy rates.[6,7] Along with this increase in clinical interest has come an increase in published literature on the topic of ankyloglossia.[8] As is the case for any rapid change in medicine, high-quality research has often lagged behind the explosion of clinical interest, leaving clinicians with little guidance regarding the best approach for diagnosis and treatment, and generating heated controversy in the medical community.[9,10]

The incidence of tongue-tie in the general population is estimated between 0.02% and 12%. These rates are variable, at least in part, because no consensus exists on the best way to diagnose tongue-tie.[11] Using the Coryllos classification (see later discussion), Haham and colleagues[12] evaluated 200 newborns within 3 days of birth, and found that all but 1 baby had “an observable or palpable lingual frenulum” that was Coryllos class 1 (frenulum attached to the tip of tongue) through 4 (frenulum attaches at the base of tongue). The investigators found no correlation between Coryllos type and presence of breastfeeding difficulty. Two studies using the Hazelbaker tool

reported an incidence and prevalence ranging from 4.2% to 12.8%.[13,14] Other studies attempting to estimate the incidence and prevalence of tongue-tie in babies have used varying methods for diagnosis, including "when the lingual frenulum attaches close to the tongue tip," "when the frenum extends to the papillary surface of the tongue," and "when the frenum caused a fissure in the tongue during movement."[11,12] Characterization of posterior tongue-tie has proven even more difficult and controversial. Descriptions of posterior tongue-tie include the presence of a "thick, fibrous cord posterior to the ventral tongue mucosa…obscured by the 'mucosal curtain'"[15] or "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."[16] In an observational study, up to 59% of healthy newborns fit these criteria.[17] The true incidence of posterior tongue-tie is unknown.[18] The natural history and outcomes for patients with tongue-tie also have not been established.[19] An online Web-based survey conducted by Jin and colleagues[19] found that professional opinions about tongue-tie varied greatly based on profession and geography. One recent study looked at Website quality and trends for ankyloglossia, and found that, although overall the quality of Websites is good, many of the published Websites available to patients and the community are opinion pieces without clear sources and with inherent bias toward performing frenotomy for tongue-tie.[20] Potentially in response to the marked increase in the diagnosis and treatment of tongue-tie, some centers have put safeguards in place to minimize unnecessary procedures in newborns thought to have tongue-tie.[21]

As understanding of ankyloglossia and other oral ties evolves, the epidemiology of this disorder will continue to change as well (**Fig. 1**).

## GENETICS

Multiple studies consistently demonstrate male-to-female ratios from 1.1:1 to 3:1.[14,22–34] These findings would suggest X-linked or autosomal dominant inheritance. This understanding is complicated by distinctions between sporadic and familial cases of ankyloglossia.[35] Most cases of ankyloglossia are thought to be sporadic and have a higher male predilection than the familial cases. Environmental or teratogen causes of ankyloglossia have been reported as well.[36] Additional heterogeneity is seen with differing ankyloglossia grading types. O'Callahan and colleagues[37] reported that the male predominance decreased from 68% for Coryllos types 1 and 2, to 59% for type 3, and to 46% for type 4 ankyloglossia. Similar trends were noted by Haham and colleagues.[12]

An X-linked cause is supported by the X-linked cleft palate syndrome.[22,38] This syndrome is caused by TBX22 gene mutation, a T-box gene involved in early vertebrate development. In addition to this association, in isolated ankyloglossia cases, a Finnish familial cohort demonstrated autosomal dominant inheritance with no TBX22 mutations.[23,24] Another pedigree study of 149 Korean subjects with isolated ankyloglossia was most suggestive of X-linked inheritance.[39] A mouse model of LGR5 knockout, a G protein–coupled receptor, has also been demonstrated to be a candidate gene for ankyloglossia in mice.[25] The mice have high mortality but do consistently have ankyloglossia. No studies have demonstrated LGR5 mutations in humans with ankyloglossia.

An autosomal dominant inheritance has been suggested through pedigree studies of families with inherited ankyloglossia.[35] Studies that evaluate multiple generations of familial ankyloglossia follow an autosomal dominant with incomplete penetrance pattern rather than X-linked.[23,26,35,39] In these situations, both TBX22 and LGR5 were excluded as potential candidates.

**Fig. 1.** Coryllos Lingual Frenulum Classification. Top to bottom: type 1 attaches to tip of tongue, type 2 attaches 2-4 mm from tongue tip, type 3 attaches to mid-tongue, type 4 attaches with a posterior attachment.

With these findings in mind, the embryologic development of the tongue is still an evolving story. The described inheritance patterns found in ankyloglossia are consistent with significant genetic heterogeneity with no clear single gene locus currently known.

## PRESENTATION AND IMPACT

Tongue-tie in infants and children can have wide-ranging effects, depending on the age of the child and the degree and pattern of tongue-tie. The domain in which this has been studied most is the effect of tongue-tie on breastfeeding in infants. Effective breastfeeding requires formation of an adequate seal between the baby's oral cavity and the mother's nipple, which allows creation of an intraoral vacuum, and a coordinated peristaltic mechanism. Tongue position is important for all of these steps.[3,40] Some of this positioning and suckling can be observed directly, paying special attention to the depth of the baby's latch, adequate positioning, sucking motion, and coordination. Ultrasound studies have also been helpful in characterizing the mechanism of breastfeeding, including the intraoral positioning and motions of the tongue.[41]

For babies noted to have tethering of the tongue, some characteristic presentation features have been widely recognized, whereas others are still being debated. Consensus in the literature and from clinical experience suggests that babies with limited tongue mobility due to ankyloglossia can have a shallow latch and poor oral seal around the nipple. This can result in maternal nipple pain, which is sometimes severe enough to cause early cessation of breastfeeding. Nipple trauma such as cracking, bleeding, and ulceration can also occur, leading to plugged milk ducts and mastitis in severe cases. Puapornpong and colleagues[42] studied nipple pain specifically in 1649 postpartum breastfeeding women. They found that moderate to severe tongue-tie was the primary reason for nipple pain in 122 women and that frenotomy was effective in relieving this pain. Ankyloglossia also makes the transfer of milk less efficient, which can lead to a decrease in milk supply, poor infant weight gain, prolonged feedings, and failure to thrive. The intermittent loss of the oral nipple seal also leads to a clicking sound and aerophagia, which some investigators have hypothesized may contribute to symptomatic reflux in infants.[43,44] The psychosocial wellbeing of both mother and child can be affected by a short lingual frenulum, in particular for mothers who are unable to continue breastfeeding. Wong and colleagues[45] looked at online forum discussions among breastfeeding women and found that many threads contained language related to maternal distress, including fear, anger, frustration, and depressed mood.

Tethering of the tongue from ankyloglossia also raises concerns about oral hygiene, dental health, and orthognathic development. Tongue mobility is essential for sweeping the oral cavity and cleaning the teeth after feedings, to remove residual food debris that could contribute to caries. An association between a tight lingual frenulum and increased dental caries is thought to be due to restrictions in the ability of the tongue to adequately reach and clean all of the teeth on a regular basis. In addition to dental caries, some studies have implicated the lingual frenulum in contributing to problems with normal occlusion and craniofacial development.[46–48] This is based on the premise that alterations in tongue mobility, sucking motion, and coordination can alter the developmental balance of the stomatognathic system, leading to changes in the shape of the dental arches and malocclusion.[48] Specifically, 2 studies have reported an association between a short lingual frenulum and class III occlusion.[46,47] In the absence of larger prospective studies, establishing a causal link is more challenging.

There is evidence in the literature that ankyloglossia can also have an impact on speech articulation disorders. Messner and Lalakea[49] conducted a prospective study

evaluating speech articulation in 30 children before and after frenuloplasty. There were documented articulation errors thought to be due to tongue-tie in 15 out of 21 subjects, 9 of whom improved following frenuloplasty. In addition to improvement on speech pathology reports, the parents' perception of speech intelligibility also improved significantly. Dollberg and colleagues[50] looked at speech intelligibility in children 4 to 8 years old who had difficulty with tongue-tie as infants. Half of these infants had been treated with frenulectomy and half with observation. The investigators found that intelligibility was not different between children with untreated tongue-tie versus those with treated tongue-tie. There was no comparison with a control group without tongue-tie. A study evaluating speech outcomes for subjects older than the age of 3 years undergoing frenulum Z-plasty found that a 4-flap frenulum Z-plasty resulted in greater improvements in articulation compared with a horizontal-to-vertical Z-plasty.[51] Although some studies lend support to the idea that tethering of the tongue affects speech articulation in some children with ankyloglossia, and that releasing the frenulum has the potential to create objective and subjective improvements in speech intelligibility, the evidence is limited and sometimes contradictory. A comprehensive review by Webb and colleagues[52] concluded that the quality of evidence regarding speech outcomes is low and definite conclusions cannot be drawn. Most clinicians advise against performing frenotomy or frenuloplasty in a newborn infant for an indication of concerns about speech disorders in the future because there is currently no reliable way to predict which of these babies will go on to have speech problems.

Ankyloglossia most often presents in isolated fashion in familial reports, but it can also appear in syndromic form, such as in Ehlers Danlos and oro-facio-digital syndrome.[53–55] Male-to-male transmission in some families supports an autosomal dominant inheritance pattern, with incomplete penetrance.[26,35] Other genes implicated in ankyloglossia show an X-linked pattern, as with mutations in TBX22, also associated with cleft palate.[38] The lingual frenulum was also reported to be abnormal as part of the spectrum of birth defects associated with congenital Zika syndrome, either as an absent lingual frenulum[56] or as a posteriorly positioned frenulum.[57]

There several other functional and social considerations related to ankyloglossia that may need to be considered. School-aged children may be more bothered by limitations in tongue elevation and protrusion that can lead to difficulty participating in social events, including licking an ice cream cone and kissing in teens.[4] Some students have reported difficulty playing wind instruments due to poor tongue positioning. Adolescents can also struggle wearing orthodontic and dental appliances because the lingual frenulum interferes with comfortable insertion. Although these concerns are often not mentioned in the scientific literature, they are heard commonly in providers' offices and can be primary reasons for older children and their families to seek treatment of release of the lingual frenum.

## DIAGNOSTICS

Universally agreed on classification and consistent diagnostic algorithms are lacking for the diagnosis of ankyloglossia. The severe form of ankyloglossia is readily apparent, it is for mild to moderate ankyloglossia that confusion arises. Diagnosis not only uses patient and maternal symptoms but also requires a thorough oral cavity structural and functional examination. A brief visual inspection of the tongue is inadequate for diagnosis. Current understanding of ankyloglossia and its impact on breastfeeding necessitates anatomic and functional grading.

With regard to diagnostic examination, the initial examination can be performed in infants lying supine with their head toward the examiner. The mouth and lips are

examined, the resting tongue position is noted, and tongue and suck reflexes are elicited. Craniofacial appearance is noted along with jaw size and position, nasal airway, and retrognathia. The tongue is then elevated with fingers or a grooved retractor. The authors find that a grooved retractor may alter the tongue appearance and the frenulum may be missed or overestimated depending on tongue and retractor position. Attachment location on the tongue and alveolus, frenulum length, and thickness is evaluated. After anatomic assessment, a gloved finger is used to illicit a suck reflex. The latch functions, such as cupping, lift, peristalsis, suction, and snap back, can then be evaluated. Additional assessment can be made with a breastfeeding or bottle feeding trial.

Most grading systems for anatomic criteria use the point of tongue attachment, length of the frenulum, or tongue protrusion.[49,58–60] The Coryllos classification has 4 types of frenulum based on the point of attachment and is the most widely used.[58] Similar to Coryllos system, the Kotlow grading systems measure the free tongue length from the tip of the tongue to the frenulum attachment.[59,60] The frenulum length, as measured from origin to insertion, interincisal distance, or tongue protrusion, are not often practical in infants or impossible to obtain owing to lack of erupted dentition.[61–64] Functional anatomy classifications include the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) and the Bristol Tongue Assessment Tool (BTAT).[65,66] The HATLFF is a 12-item scale with 10 points for frenulum appearance and 14 points for tongue function. A score of 24 has normal function and normal anatomy. According to the scale, frenotomy is necessary for symptomatic ankyloglossia if the appearance score is less than 8 or the function score is less than 11. For perfect function scores regardless of appearance, frenotomy is not recommended. By allowing for functional assessment, it enables inclusion of posterior ankyloglossia, which may be missed in attachment-only scales. Routine use of the HATLFF can be difficult because assessment is complex and it requires an in-depth understanding of function and anatomy.[27,67] The BTAT was developed to incorporate much of the benefits of the HATLFF but to make it more portable and teachable. The scale has 4 items to grade tongue tip appearance, alveolar attachment location, tongue lift, and tongue protrusion. It was found to correlate well with HATLFF findings.[66] However, it does not fully address functional assessment. More recent studies have incorporated breastfeeding ultrasound to examine latch and tongue position, as well as milk intake.[28] Breastfeeding ultrasound is a promising tool can assist with functional assessment but is not specific to ankyloglossia.

In summary, current best practice should include an objective standardized anatomic and functional assessment of tongue appearance and mobility, as well as latch, regardless of grading system used.

## TREATMENT

There are a variety of intervention options for breastfeeding difficulty in infants. These include changes in breastfeeding technique or positioning, or assisting devices such as nipple shields. Other complementary medical treatments have included craniosacral therapy, orofacial myofunctional therapy, chiropractic care, and naturopathy. Many of these same treatment options have been used to improve breastfeeding in the setting of suspected ankyloglossia. There are no high-quality outcome studies to inform providers on the efficacy of these techniques.

Other than lactation consultant expertise and assistance, the primary treatment of ankyloglossia in infants is frenotomy, which involves surgical division of the lingual frenum. Additional surgical techniques such as frenulectomy, or removal of the frenulum; frenotomy with myotomy; and frenulum Z-plasty have been

described.[51,61,68–70] Despite these additional surgical options, frenotomy is sufficient in most infants. Shared decision-making about treatment options, with involvement of the family, is encouraged (Fig. 2).

The frenotomy technique isolates the frenulum with a grooved retractor. The frenulum is divided through the fibroelastic tissues of the frenum but not into the muscle, with scissors. Hemostasis is typically achieved with digital pressure, gauze with oxymetazoline, silver nitrate, or (rarely) suturing. Other common techniques follow the same principles but use carbon dioxide, diode, erbium: yttrium aluminium garnet

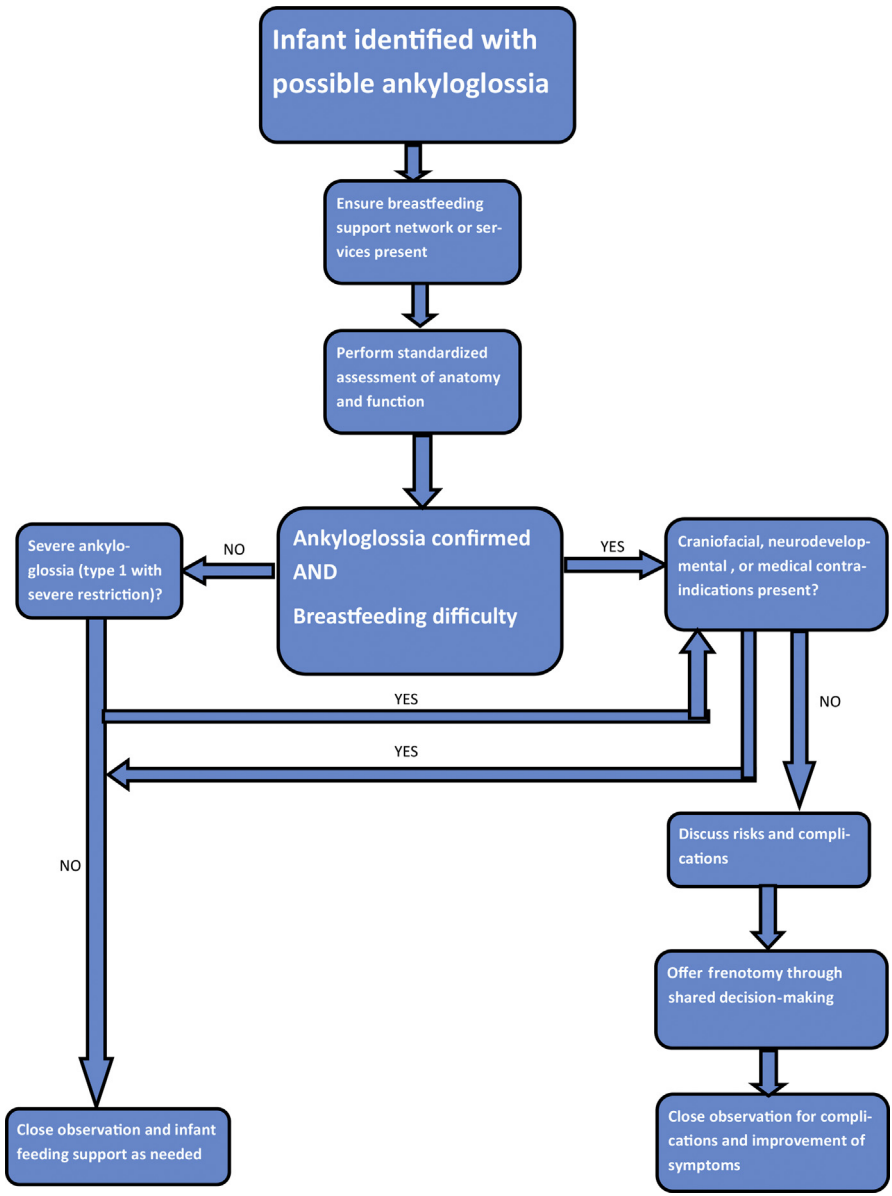


**Fig. 2.** Ankyloglossia flow chart.

(YAG), or neodymium-doped (Nd):YAG lasers, or electrocautery.[60,68,71–73] There are no comparative trials of scissor versus laser techniques for frenotomy in infants; however, in animal models of oral injury, cold surgical techniques have quicker healing,[74,75] possibly due to reduced thermal injury to the frenum and surrounding tissues. One study of adult labial frenotomy showed slight benefit of diode laser,[76] with an added benefit of concurrent hemostasis.

With regard to topical anesthesia, a randomized clinical trial concluded that topical anesthesia options are ineffective and need not be used.[77] An additional risk of topical anesthetic agents is methemoglobinemia. Common topical amide and ether anesthetics such as benzocaine have been implicated, and children younger the age of 2 years are at particular risk.[78] Instead of topical anesthesia, a 24% sucrose solution given orally before the procedure, accompanied by postprocedure nutritive or nonnutritive sucking, can help reduce discomfort.[79]

## OUTCOMES

As previously discussed, a variety of dental, speech, feeding, and sleep-related outcomes are attributed to untreated ankyloglossia. Unfortunately, the ability to predict the natural history of untreated ankyloglossia is limited owing to the wide range of published ages of presentation, literature bias toward treatment, wide variation of symptom severity, and difficulty determining causation from retrospective association studies. Two systematic reviews recently analyzed the published studies regarding the impact of surgical treatment of ankyloglossia on breastfeeding and nonbreastfeeding issues. These reports noted improvement in breastfeeding and nipple pain as assessed by maternal report, but the overall strength and quality of evidence in support of frenotomy is low.[80,81] Outside of surgical complications, surgical frenotomy outcomes are consistently positive. Most outcomes are prospective cohort or retrospective studies. Of the few published randomized controlled trials (RCTs), most of the control subjects had crossover to frenotomy as part of the study designs. Two RCTs evaluated immediate results only because most control subjects had frenotomy shortly after sham procedure.[82,83] Three other RCTs had almost entire crossover of the control arms to the treatment groups during the course of the study.[84–86]

It is the inherent bias and heterogeneity within the literature that makes comprehensive assessment of frenotomy outcomes difficult. What is clear from the body of evidence is that some infants will indeed benefit from frenotomy and others who have multifactorial causes of feeding difficulty may have minimal or no benefit from the procedure. The positive predictive value of preoperative diagnosis is limited compared with rising frenotomy treatment.[5] The need for revision treatment of recurrent symptoms has not been studied directly, but rates for revision have been reported between 2.6% and 6.5%.[87,88] For posterior tongue-tie, the revision rates can be as high as 21%.[16]

Expected treatment outcomes change by the type of ankyloglossia, that is, anterior or posterior ankyloglossia, and presence of associated findings, such as ULT. A recent Web-based postintervention maternal report study found 100% improvement in latching difficulties for type 1 and type 2 classic ankyloglossia, and 49% improvement for type 4 posterior ankyloglossia. For maternal pain, there was 79% improvement in type 1 and 2, and 63% improvement in type 4.[37] The maternal breast's transition to breastfeeding or the experience of the mother may also play a role beyond the presence of ankyloglossia.[17] First-time mothers seem to be more likely to have an infant who is diagnosed with ankyloglossia and are also less likely determine if frenotomy had been performed based on breastfeeding efficacy.[5,85]

Despite these limitations, treatment outcome measures, such as the Breastfeeding Self-Efficacy Scale (BSES), the Infant Breastfeeding Assessment Tool (IBFAT), and the Latch, Audible swallowing, Type of nipple, Comfort, Hold (LATCH) tool have generally shown improvements in retrospective studies, but controlled studies are limited.[80]

There are insufficient data to determine the association of ankyloglossia with issues other than breastfeeding.[5182] Therefore, the effect of frenotomy on speech, malocclusion, mandibular incisor irregularity, gingival recession, mandibular growth, and tongue mobility in older children is primarily described only in retrospective or cohort studies. Because tongue mobility can be restricted in ankyloglossia, the restricted mobility may affect speech articulation.[89] Messner and Lalakea[49] described the improvement of speech articulation issues in a cohort of children with frenotomy. The improvement or prevention of dental and craniofacial abnormalities is controversial, with benefits shown only in small retrospective case series.[55]

## POTENTIAL COMPLICATIONS

Frenotomy is considered to be a safe procedure in almost all cases. The most common risks include infection and minor bleeding at the site, pain and discomfort, and a risk of injury to the salivary ducts that are located near the frenulum. Other considerations that should be discussed with patients and family members during informed consent include the possibility of no improvement or the need for additional revision surgical procedures in the future. Pain and discomfort associated with the procedure are often concerns because topical anesthetic agents are contraindicated in children younger than the age of 2 years, who are at higher risk of methemoglobinemia (www.fda.gov/Drugs/DrugSafety/ucm608265.htm). Postprocedure pain from extensive dissection or cautery in the floor of mouth can lead to oral aversion in severe cases. Most research studies that have been done on tongue-tie report no complications occurring in their cohort of subjects,[82,84,85] thus lending weight to the idea that frenotomy is, in most cases, without significant morbidity.

Despite this common understanding among providers and the public about the relative safety of frenotomy, there have been case reports of very serious complications occurring after frenotomy. As the diagnosis and treatment of tongue-tie have increased in the past 20 years (see previous discussion), these cautionary reports began to appear in the literature. One of the earliest of such case reports was published by Isaiah and Pereira[90] in 2013, regarding a 2-year-old boy who underwent outpatient frenotomy under general anesthesia with scissor technique and presented on postoperative day 2 with fever, tongue swelling, and drooling. Intraoral examination revealed an infected sublingual hematoma. This patient was treated with local evacuation and debridement, intravenous steroids, and antibiotics, and he made a full recovery.

Another case report included 2 patients with Pierre Robin sequence who presented with feeding difficulty and a short lingual frenulum.[91] Both patients developed increasing respiratory symptoms after frenotomy, presumed to be due to worsening of glossoptosis after the anterior attachments of the tongue to the floor of mouth had been released. Both patients had significant daytime symptoms of cyanosis and increased work of breathing with feeds, and both had severe obstructive sleep apnea noted on overnight polysomnogram. Surgical management of glossoptosis (tongue–lip adhesion in 1 patient and mandibular distraction osteogenesis in the other) was effective in improving respiratory symptoms; however, both patients eventually required feeding tube insertion for inadequate oral intake.

In 2017, Tracy and colleagues[92] described 2 pediatric patients who experienced heavy intraoral bleeding leading to hypovolemic shock. Both patients were older

than 1 year of age and underwent frenotomy under anesthesia or sedation, and both presented hours to days after the procedure with significant intraoral bleeding requiring transfusion in both cases and cardiopulmonary resuscitation in 1 case.

These reports highlight the risk of airway compromise with any complication arising in the floor of mouth. Otolaryngologists are particularly sensitive to airway compromise from infection or hematoma in the floor of mouth, but other practitioners who also perform frenotomy may not be as attuned to this possibility and may not be comfortable or facile in managing the airway in these rare circumstances. All providers treating tongue-tie need to be aware that such serious complications can occur so that patients receive timely identification and treatment.

## MAXILLARY AND OTHER ORAL TIES

Along with the increased interest in lingual ties, there has been a recent increase in clinical concern about labial ties, including the maxillary and mandibular midline frena, as well as lateral oral ties. To date, the literature on the anatomic significance and potential impact of these ties is sparse. Cadaver studies have demonstrated that these frena are composed of dense collagenous connective tissue, often at the reflections or interface of musculature in the area.[93,94] Observational studies have shown a wide variety of morphologic variation, with some investigators advising caution about interpreting these normal variants as pathologic.[95] Many potential issues have been purported to be due to tight maxillary and mandibular labial ties, including formation of a midline diastema between the teeth, increased caries and periodontal disease, gingival recession, difficulty in wearing dentures or retainers, difficulty with lip mobility, and possible esthetic or psychological consequences.[93,94]

The midline maxillary labial frenum, which can form an ULT, has recently been implicated in breastfeeding difficulty due to inability to properly flange the upper lip to obtain a proper latch. Because of the controversy surrounding the identification, diagnosis, and impact of an ULT, recent studies have attempted to characterize the normal anatomy of the maxillary labial frenum in newborns.[96,97] Santa Maria and colleagues[97] studied 100 healthy newborns and developed the Stanford Classification System, which had higher interrater and intrarater reliability than prior studies. Up to 11% of these babies had a class 3 upper lip frenulum that attached along the inferior margin of the alveolar papilla or onto the palate. The authors point out that the natural history of this structure is not known, and performing procedures to release it without adequate evidence that it is impactful may not be advisable.

Clinical research on tongue-tie may also include subsets of subjects with both tongue-tie and ULT. Pransky and colleagues[98] reported that, in a group of 618 infants with breastfeeding difficulty, 2% had an isolated ULT, 6% had concurrent ULT and anterior ankyloglossia, and 5% had concurrent ULT and posterior ankyloglossia. With treatment of these ties, more than 85% to 100% reported improvement in breastfeeding. Of those with isolated ULT, 100% reported improvement in breastfeeding after release. Benoiton and colleagues[99] also reported on a mixed group of subjects with tongue-tie and ULT and found that, for isolated ULT, 2 out of 3 subjects reported improvement, and, for combined posterior tongue-tie and ULT, 9 out of 10 subjects eventually reported improvement in breastfeeding. Finally, in 2017, Ghaheri and colleagues[44] reported a statistically significant improvement on the BSES following release of both tongue-tie and ULT in a cohort of 178 infants. Results from these studies are difficult to interpret due to various methodological limitations, including a lack of control group, assessment of procedures performed in combination, and

subjective outcome measures. More evidence will be needed before division of ULT or other oral ties can be recommended for breastfeeding difficulty.

## SUMMARY

Ankyloglossia is a complex multidisciplinary issue, with interplay and impact on a mother–infant dyad, and with many providers from different backgrounds involved in evaluation and treatment. Knowledge about the genetics and epidemiology of this group of disorders continues to evolve. To date, there is high-quality evidence from randomized prospective trials that division of an anterior tongue-tie is helpful in improving breastfeeding in some dyads. Although evidence is emerging on posterior tongue-tie and other oral ties, these studies are often limited by a lack of consensus on diagnosis, indications for treatment, and validated outcomes. As understanding improves, all disciplines will need to work toward cohesive treatment recommendations based on high-quality evidence.

## REFERENCES

1. Knox I. Tongue tie and frenotomy in the breastfeeding newborn. Neoreviews 2010;11:e513–9.
2. Obladen M. Much ado about nothing: two millennia of controversy on tongue-tie. Neonatology 2010;97:83–9.
3. Hall DMB, Renfrew MJ. Tongue tie. Arch Dis Child 2005;90:1217–8.
4. Messner AH, Lalakea ML. Ankyloglossia: controversies in management. Int J Pediatr Otorhinolaryngol 2000;54(2–3):123–31.
5. Walsh J, Links A, Boss E, et al. Ankyloglossia and lingual frenotomy: national trends in inpatient diagnosis and management in the United States, 1997-2012. Otolaryngol Head Neck Surg 2017;156(4):735–40.
6. Lisonek M, Liu S, Dzakpasu S, et al. Changes in the incidence and surgical treatment of ankyloglossia in Canada. Paediatr Child Health 2017;22(7):382–6.
7. Kapoor V, Douglas PS, Hill PS, et al. Frenotomy for tongue-tie in Australian children, 2006-2016: an increasing problem. Med J Aust 2018;208(2):88–9.
8. Bin-Nun A, Kasirer YM, Mimouni FB. A dramatic increase in tongue tie-related articles: a 67 years systematic review. Breastfeed Med 2017;12(7):410–4.
9. Douglas PS. Conclusions of Ghaheri's Study that laser surgery for posterior tongue and lip ties improves breastfeeding are not substantiated. Breastfeed Med 2017;12:180–1.
10. Ghaheri BA, Cole M. Response to Douglas Re: "Conclusions of Ghaheri's Study that laser surgery for posterior tongue and lip ties improves breastfeeding are not substantiated. Breastfeed Med 2017;12:182–3.
11. Segal LM, Stephenson R, Dawes M, et al. Prevalence, diagnosis, and treatment of ankyloglossia: methodologic review. Can Fam Physician 2007;53(6):1027–33.
12. Haham A, Marom R, Mangel L, et al. Prevalence of breastfeeding difficulties in newborns with a lingual frenulum: a prospective cohort series. Breastfeed Med 2014;9(9):438–41.
13. Ballard JL, Auer CE, Khoury JC. Ankyloglossia: assessment, incidence, and effect of frenuloplasty on the breastfeeding dyad. Pediatrics 2002;110(5):e63.
14. Ricke LA, Baker NJ, Madlon-Kay DJ, et al. Newborn tongue-tie: prevalence and effect on breast-feeding. J Am Board Fam Pract 2005;18(1):1–7.
15. Chu MW, Bloom DC. Posterior ankyloglossia: a case report. Int J Pediatr Otorhinolaryngol 2009;73(6):881–3.

16. Hong P, Lago D, Seargeant J, et al. Defining ankyloglossia: a case series of anterior and posterior tongue ties. Int J Pediatr Otorhinolaryngol 2010;74(9):1003–6.
17. Walker RD, Messing S, Rosen-Carole C, et al. Defining tip-frenulum length for ankyloglossia and its impact on breastfeeding: a prospective cohort study. Breastfeed Med 2018;13(3):204–10.
18. Douglas PS. Rethinking "posterior" tongue-tie. Breastfeed Med 2013;8(6):503–6.
19. Jin RR, Sutcliffe A, Vento M, et al. What does the world think of ankyloglossia? Acta Paediatr 2018;107(10):1733–8.
20. Aaronson NL, Castaño JE, Simons JP, et al. Quality, readability, and trends for websites on ankyloglossia. Ann Otol Rhinol Laryngol 2018;127(7):439–44.
21. Dixon B, Gray J, Elliot N, et al. A multifaceted programme to reduce the rate of tongue-tie release surgery in newborn infants: observational study. Int J Pediatr Otorhinolaryngol 2018;113:156–63.
22. Gorski SM, Adams KJ, Birch PH, et al. Linkage analysis of X-linked cleft palate and ankyloglossia in Manitoba Mennonite and British Columbia Native kindreds. Hum Genet 1994;94(2):141–8.
23. Klockars T. Familial ankyloglossia (tongue-tie). Int J Pediatr Otorhinolaryngol 2007;71(8):1321–4.
24. Klockars T, Kyttanen S, Ellonen P. TBX22 and tongue-tie. Cleft Palate Craniofac J 2012;49(3):378–9.
25. Morita H, Mazerbourg S, Bouley DM, et al. Neonatal lethality of LGR5 null mice is associated with ankyloglossia and gastrointestinal distension. Mol Cell Biol 2004; 24(22):9736–43.
26. Lenormand A, Khonsari R, Corre P, et al. Familial autosomal dominant severe ankyloglossia with tooth abnormalities. Am J Med Genet A 2018;176(7):1614–7.
27. Madlon-Kay DJ, Ricke LA, Baker NJ, et al. Case series of 148 tongue-tied newborn babies evaluated with the assessment tool for lingual frenulum function. Midwifery 2008;24(3):353–7.
28. Douglas P, Geddes D. Practice-based interpretation of ultrasound studies leads the way to more effective clinical support and less pharmaceutical and surgical intervention for breastfeeding infants. Midwifery 2018;58:145–55.
29. Jorgenson RJ, Shapiro SD, Salinas CF, et al. Intraoral findings and anomalies in neonates. Pediatrics 1982;69(5):577–82.
30. Garcia Pola MJ, González García M, García Martín JM, et al. A study of pathology associated with short lingual frenum. ASDC J Dent Child 2002;69(1):59–62, 12.
31. Sedano HO. Congenital oral anomalies in Argentinian children. Community Dent Oral Epidemiol 1975;3(2):61–3.
32. Salem G, Holm SA, Fattah R, et al. Developmental oral anomalies among schoolchildren in Gizan region, Saudi Arabia. Community Dent Oral Epidemiol 1987; 15(3):150–1.
33. Voros-Balog T, Vincze N, Banoczy J. Prevalence of tongue lesions in Hungarian children. Oral Dis 2003;9(2):84–7.
34. Messner AH, Lalakea ML, Aby J, et al. Ankyloglossia: incidence and associated feeding difficulties. Arch Otolaryngol Head Neck Surg 2000;126(1):36–9.
35. Klockars T, Pitkaranta A. Inheritance of ankyloglossia (tongue-tie). Clin Genet 2009;75(1):98–9.
36. Harris EF, Friend GW, Tolley EA. Enhanced prevalence of ankyloglossia with maternal cocaine use. Cleft Palate Craniofac J 1992;29(1):72–6.
37. O'Callahan C, Macary S, Clemente S. The effects of office-based frenotomy for anterior and posterior ankyloglossia on breastfeeding. Int J Pediatr Otorhinolaryngol 2013;77(5):827–32.

38. Braybrook C, Doudney K, Marçano AC, et al. The T-box transcription factor gene TBX22 is mutated in X-linked cleft palate and ankyloglossia. Nat Genet 2001; 29(2):179–83.
39. Han SH, Kim MC, Choi YS, et al. A study on the genetic inheritance of ankyloglossia based on pedigree analysis. Arch Plast Surg 2012;39(4):329–32.
40. Patel J, Anthonappa RP, King NM. All tied up! influences of oral frenulae on breastfeeding and their recommended management strategies. J Clin Pediatr Dent 2018;42(6):407–13.
41. Geddes DT, Langton DB, Gollow I, et al. Frenulotomy for breastfeeding infants with ankyloglossia: effect on milk removal and sucking mechanism as imaged by ultrasound. Pediatrics 2008;122(1):e188–94.
42. Puapornpong P, Paritakul P, Suksamarnwong M, et al. Nipple pain incidence, the predisposing factors, the recovery period after care management, and the exclusive breastfeeding outcome. Breastfeed Med 2017;12:169–73.
43. Kotlow LA. The influence of the maxillary frenum on the development and pattern of dental caries on anterior teeth in breastfeeding infants: prevention, diagnosis, and treatment. J Hum Lact 2010;26(3):304–8.
44. Ghaheri BA, Cole M, Fausel SC, et al. Breastfeeding improvement following tongue-tie and lip-tie release: a prospective cohort study. Laryngoscope 2017; 127(5):1217–23.
45. Wong K, Patel P, Cohen MB, et al. Breastfeeding infants with ankyloglossia: insight into mothers' experiences. Breastfeed Med 2017;12:86–90.
46. Jang GJ, Lee SL, Kim HM. [Breast feeding rates and factors influencing breast feeding practice in late preterm infants: comparison with preterm born at less than 34 weeks of gestational age]. J Korean Acad Nurs 2012;42(2):181–9.
47. Meenakshi S, Jagannathan N. Assessment of lingual frenulum lengths in skeletal malocclusion. J Clin Diagn Res 2014;8(3):202–4.
48. Pompeia LE, Ilinsky RS, Ortolani CLF, et al. Ankyloglossia and its influence on growth and development of the stomatognathic system. Rev Paul Pediatr 2017; 35(2):216–21.
49. Messner AH, Lalakea ML. The effect of ankyloglossia on speech in children. Otolaryngol Head Neck Surg 2002;127(6):539–45.
50. Dollberg S, Manor Y, Makai E, et al. Evaluation of speech intelligibility in children with tongue-tie. Acta Paediatr 2011;100(9):e125–7.
51. Heller J, Gabbay J, O'Hara C, et al. Improved ankyloglossia correction with four-flap Z-frenuloplasty. Ann Plast Surg 2005;54(6):623–8.
52. Webb AN, Hao W, Hong P. The effect of tongue-tie division on breastfeeding and speech articulation: a systematic review. Int J Pediatr Otorhinolaryngol 2013; 77(5):635–46.
53. Devasya A, Sarpangala M. Familial ankyloglossia -a rare report of three cases in a family. J Clin Diagn Res 2017;11(2):ZJ03–4.
54. Mintz SM, Siegel MA, Seider PJ. An overview of oral frena and their association with multiple syndromic and nonsyndromic conditions. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2005;99(3):321–4.
55. Suter VG, Bornstein MM. Ankyloglossia: facts and myths in diagnosis and treatment. J Periodontol 2009;80(8):1204–19.
56. Del Campo M, Feitosa IM, Ribeiro EM, et al. The phenotypic spectrum of congenital Zika syndrome. Am J Med Genet A 2017;173(4):841–57.
57. Fonteles CSR, Marques Ribeiro E, Sales Aragão Santos M, et al. Lingual frenulum phenotypes in Brazilian infants with congenital Zika syndrome. Cleft Palate Craniofac J 2018;55(10):1391–8.

58. Breastfeeding, A.A.o.P.S.o.. Congenital tongue-tie and its impact on breastfeeding 2004. Available at: https://urldefense.proofpoint.com/v2/url?u=http-3A__www.lunalactation.com_Ankyloglossia-5FAAPnewsletter.pdf&d=DwIFAg&c=4sF48jRmVAe_CH-k9mXYXEGfSnM3bY53YSKuLUQRxhA&r=xA2HPRJy1uN8376uKwD33sQTRrSRYfMjp9TI71yIeL8&m=f-py4WBjNhKe58-8Xxm4cFfycl70Z-2t0fX2zufQ5TE&s=sw6dtLPnRKe0hpR0YtZHeVYSlBYI4ZB9ovFCiWsUx6U&e=. Accessed July 7, 2019.
59. Kotlow LA. Ankyloglossia (tongue-tie): a diagnostic and treatment quandary. Quintessence Int 1999;30(4):259–62.
60. Kotlow L. Diagnosis and treatment of ankyloglossia and tied maxillary fraenum in infants using Er:YAG and 1064 diode lasers. Eur Arch Paediatr Dent 2011;12(2):106–12.
61. Lalakea ML, Messner AH. Ankyloglossia: does it matter? Pediatr Clin North Am 2003;50(2):381–97.
62. Williams WN, Waldron CM. Assessment of lingual function when ankyloglossia (tongue-tie) is suspected. J Am Dent Assoc 1985;110(3):353–6.
63. Notestine GE. The importance of the identification of ankyloglossia (short lingual frenulum) as a cause of breastfeeding problems. J Hum Lact 1990;6(3):113–5.
64. Ruffoli R, Giambelluca MA, Scavuzzo MC, et al. Ankyloglossia: a morphofunctional investigation in children. Oral Dis 2005;11(3):170–4.
65. Hazelbaker AK. Tongue-tie morphogenesis, impact, assessment and treatment. Columbus (OH): Aidan & Eva Press; 2010.
66. Ingram J, Johnson D, Copeland M, et al. The development of a tongue assessment tool to assist with tongue-tie identification. Arch Dis Child Fetal Neonatal Ed 2015;100(4):F344–8.
67. Ngerncham S, Laohapensang M, Wongvisutdhi T, et al. Lingual frenulum and effect on breastfeeding in Thai newborn infants. Paediatr Int Child Health 2013;33(2):86–90.
68. Junqueira MA, Cunha NN, Costa e Silva LL, et al. Surgical techniques for the treatment of ankyloglossia in children: a case series. J Appl Oral Sci 2014;22(3):241–8.
69. Choi YS, Lim JS, Han KT, et al. Ankyloglossia correction: Z-plasty combined with genioglossus myotomy. J Craniofac Surg 2011;22(6):2238–40.
70. Horton CE, Crawford HH, Adamson JE, et al. Tongue-tie. Cleft Palate J 1969;6:8–23.
71. Aras MH, Göregen M, Güngörmüş M, et al. Comparison of diode laser and Er:-YAG lasers in the treatment of ankyloglossia. Photomed Laser Surg 2010;28(2):173–7.
72. Tuli A, Singh A. Monopolar diathermy used for correction of ankyloglossia. J Indian Soc Pedod Prev Dent 2010;28(2):130–3.
73. Chiniforush N, Ghadimi S, Yarahmadi N, et al. Treatment of ankyloglossia with carbon dioxide (CO2) laser in a pediatric patient. J Lasers Med Sci 2013;4(1):53–5.
74. Morosolli AR, Veeck EB, Niccoli-Filho W, et al. Healing process after surgical treatment with scalpel, electrocautery and laser radiation: histomorphologic and histomorphometric analysis. Lasers Med Sci 2010;25(1):93–100.
75. D'Arcangelo C, Di Nardo Di Maio F, Prosperi GD, et al. A preliminary study of healing of diode laser versus scalpel incisions in rat oral tissue: a comparison of clinical, histological, and immunohistochemical results. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2007;103:764–73.
76. Gandhi D, Gandhi P. Comparison of healing period after frenectomy using scalpel, electrocautery and diode laser. Br J Med Med Res 2017;21(12):1–9.

77. Shavit I, Peri-Front Y, Rosen-Walther A, et al. A randomized trial to evaluate the effect of two topical anesthetics on pain response during frenotomy in young infants. Pain Med 2017;18(2):356–62.
78. Avarello JT, Gupta A, Silverman RA. Post-frenotomy methemoglobinemia associated with mepivacaine use in a 3 day old. Emergency Med 2013;3(1):1–3.
79. Stevens B, Yamada J, Ohlsson A, et al. Sucrose for analgesia in newborn infants undergoing painful procedures. Cochrane Database Syst Rev 2016;(7):CD001069.
80. Francis DO, Krishnaswami S, McPheeters M. Treatment of ankyloglossia and breastfeeding outcomes: a systematic review. Pediatrics 2015;135(6):e1458–66.
81. Chinnadurai S, Francis DO, Epstein RA, et al. Treatment of ankyloglossia for reasons other than breastfeeding: a systematic review. Pediatrics 2015;135(6):e1467–74.
82. Dollberg S, Botzer E, Grunis E, et al. Immediate nipple pain relief after frenotomy in breast-fed infants with ankyloglossia: a randomized, prospective study. J Pediatr Surg 2006;41(9):1598–600.
83. Berry J, Griffiths M, Westcott C. A double-blind, randomized, controlled trial of tongue-tie division and its immediate effect on breastfeeding. Breastfeed Med 2012;7(3):189–93.
84. Hogan M, Westcott C, Griffiths M. Randomized, controlled trial of division of tongue-tie in infants with feeding problems. J Paediatr Child Health 2005;41(5–6):246–50.
85. Buryk M, Bloom D, Shope T. Efficacy of neonatal release of ankyloglossia: a randomized trial. Pediatrics 2011;128(2):280–8.
86. Emond A, Ingram J, Johnson D, et al. Randomised controlled trial of early frenotomy in breastfed infants with mild-moderate tongue-tie. Arch Dis Child Fetal Neonatal Ed 2014;99(3):F189–95.
87. Argiris K, Vasani S, Wong G, et al. Audit of tongue-tie division in neonates with breastfeeding difficulties: how we do it. Clin Otolaryngol 2011;36(3):256–60.
88. Steehler MW, Steehler MK, Harley EH. A retrospective review of frenotomy in neonates and infants with feeding difficulties. Int J Pediatr Otorhinolaryngol 2012;76(9):1236–40.
89. Lalakea ML, Messner AH. Ankyloglossia: the adolescent and adult perspective. Otolaryngol Head Neck Surg 2003;128(5):746–52.
90. Isaiah A, Pereira KD. Infected sublingual hematoma: a rare complication of frenulectomy. Ear Nose Throat J 2013;92(7):296–7.
91. Genther DJ, Skinner ML, Bailey PJ, et al. Airway obstruction after lingual frenulectomy in two infants with Pierre-Robin sequence. Int J Pediatr Otorhinolaryngol 2015;79(9):1592–4.
92. Tracy LF, Gomez G, Overton LJ, et al. Hypovolemic shock after labial and lingual frenulectomy: a report of two cases. Int J Pediatr Otorhinolaryngol 2017;100:223–4.
93. Gartner LP, Schein D. The superior labial frenum: a histologic observation. Quintessence Int 1991;22(6):443–5.
94. Iwanaga J, Takeuchi N, Oskouian RJ, et al. Clinical anatomy of the frenulum of the oral vestibule. Cureus 2017;9(6):e1410.
95. Dasgupta P, Kamath G, Hs S, et al. Morphological variations of median maxillary labial frenum: a clinical study. J Stomatol Oral Maxillofac Surg 2017;118(6):337–41.
96. Kotlow LA. Diagnosing and understanding the maxillary lip-tie (superior labial, the maxillary labial frenum) as it relates to breastfeeding. J Hum Lact 2013;29(4):458–64.

97. Santa Maria C, Aby J, Truong MT, et al. The superior labial frenulum in newborns: what is normal? Glob Pediatr Health 2017;4. 2333794X17718896.
98. Pransky SM, Lago D, Hong P. Breastfeeding difficulties and oral cavity anomalies: the influence of posterior ankyloglossia and upper-lip ties. Int J Pediatr Otorhinolaryngol 2015;79(10):1714–7.
99. Benoiton L, Morgan M, Baguley K. Management of posterior ankyloglossia and upper lip ties in a tertiary otolaryngology outpatient clinic. Int J Pediatr Otorhinolaryngol 2016;88:13–6.