Ankyloglossia and breastfeeding

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Ankyloglossia ('tongue-tie') is a relatively common congenital anomaly characterized by an abnormally short lingual frenulum, which may restrict tongue tip mobility. There is considerable controversy regarding its diagnosis, clinical significance and management, and there is wide variation in practice in this regard. Most infants with ankyloglossia are asymptomatic and do not exhibit feeding problems. Based on available evidence, frenotomy cannot be recommended for all infants with ankyloglossia. There may be an association between ankyloglossia and significant breastfeeding difficulties in some infants. This subset of infants may benefit from frenotomy (the surgical division of the lingual frenulum). When an association between significant tongue-tie and major breastfeeding problems is clearly identified and surgical intervention is deemed to be necessary, frenotomy should be performed by a clinician experienced with the procedure and using appropriate analgesia. More definitive recommendations regarding the management of tongue-tie in infants await clear diagnostic criteria and appropriately designed trials.

Key Words: *Ankyloglossia; Breastfeeding; Frenotomy; Infant; Tongue-tie*

L'ankyloglossie et l'allaitement

L'ankyloglossie est une anomalie congénitale relativement courante caractérisée par un frein lingual anormalement court, qui peut restreindre la mobilité de la langue. Une énorme controverse en entoure le diagnostic, la signification clinique et la prise en charge, et les pratiques varient considérablement à cet égard. La plupart des nourrissons ayant une ankyloglossie sont asymptomatiques et n'ont pas de problèmes pour se nourrir. D'après les données probantes, on ne peut pas recommander la frénotomie pour tous les nourrissons ayant une ankyloglossie. Chez certains nourrissons, on peut constater une association entre l'ankyloglossie et certains problèmes d'allaitement. Ce sous-groupe de nourrissons peut tirer profit de la frénotomie (la division chirurgicale du frein de la langue). Lorsqu'une association entre une ankyloglossie marquée et de graves troubles d'allaitement est clairement établie et qu'une intervention chirurgicale est jugée nécessaire, un clinicien habitué à la frénotomie doit effectuer l'intervention et utiliser l'analgésie convenable. Pour faire des recommandations plus définitives sur la prise en charge de l'ankyloglossie chez les nourrissons, il faudra attendre des critères diagnostiques clairs et des essais cliniques bien conçus.

BACKGROUND

Ankyloglossia ('tongue-tie') is a congenital anomaly observed in newborns and children, and is characterized by an abnormally short lingual frenulum. The tight frenulum can cause decreased tongue mobility to varying degrees.

Associations between tongue-tie, lactation problems, speech disorders and other oral motor disorders (eg, problems with swallowing or licking) have been inconsistent, and are an ongoing source of controversy within the medical community.(1-3) One survey of otolaryngologists, paediatricians, speech pathologists and lactation consultants reported significant disparities within and among these groups with regard to their approaches to ankyloglossia and their beliefs regarding its association with feeding, speech and social problems.(1) Dentists are similarly divided on the topic.(4)

With a renewed emphasis on the benefits of breastfeeding (which the Canadian Paediatric Society fully supports), there is more pressure to diagnose ankyloglossia as a barrier to successful breastfeeding, thus increasing the demand for frenotomy.

The present statement specifically focuses on the evidence surrounding the association of ankyloglossia and breastfeeding difficulties in infants, and the efficacy of frenotomy in the context of ankyloglossia and breastfeeding difficulties. The present statement replaces the previous Canadian Paediatric Society document revised in 2011.(5) Several studies have been published in the interim.

DEFINITION

There is neither a universally accepted definition of ankyloglossia nor practical objective criteria for diagnosing this condition. Historically, definitions have been based on either anatomical characteristics of the lingual frenulum (ie, the degree of fusion between the child's tongue and the floor of the mouth) or on functional impairment (ie, an inability to protrude the tongue past the incisal edge of the lower gingiva and other signs of decreased tongue mobility).(1,6-9) In one classification system, ankyloglossia (Types I and II) is characterized by insertion at the tip of the tongue (Type I) or slightly behind the tip (Type II), while posterior ankyloglossia is characterized by a thickened frenulum (Type III) or a submucosal frenulum (a flat, broad mound) that restricts movement at the base of the tongue (Type IV).(10) These definitions are seldom used in the literature and rarely in the clinical domain. Hazelbaker(11) developed a descriptive assessment tool for lingual frenulum function; however, it is complex, lengthy and has not been validated in a controlled manner.(12) Criteria used to diagnose ankyloglossia show considerable variation, and there is no accepted standard. The lack of a consistent definition further fuels controversy regarding this condition and its clinical significance.

ETIOLOGY

The tongue is fused to the floor of the mouth in early development. Cell death and resorption free the tongue, with the frenulum left

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as the only remnant of initial attachment. The lingual frenulum typically becomes less prominent as a natural process of the child's growth and development, when the alveolar ridge grows in height and the teeth begin to erupt.(2) This process occurs during the first six months to five years of life. Ankyloglossia can be classified based on the degree of fusion remaining between the tongue and the floor of the mouth.(2)

There may be a genetic predisposition to ankyloglossia.(13) This congenital anomaly typically occurs in isolation.

PREVALENCE

The reported prevalence of ankyloglossia in infants is variable in the literature, reflecting the lack of a consistent definition. Estimates range from 4.2% to 10.7% in newborns.(7-9)

PATHOPHYSIOLOGY

The role of a short lingual frenulum as a cause of breastfeeding difficulties has been described in multiple anecdotal reports linking ankyloglossia to poor latch, maternal nipple pain and trauma, suboptimal infant weight gain, infant breast refusal and low maternal milk supply due to poor milk removal.(14-17) To nurse successfully, an infant must latch on to the areola using the upper gum ridge, buccal fatty pads and tongue. Suckling begins with forward movement of the jaw and tongue. The tongue helps to make a better seal, but with minimal action. The anterior edge of the tongue thins, cupping upward to begin a peristaltic ripple back toward the throat. At the same time, the lower jaw squeezes milk from the ductules.(18) It is clear that restriction of tongue movement must be extreme to interfere with sucking and swallowing.(2) It also appears that some mothers have particular breast, nipple or milk ejection characteristics that allow them to successfully breastfeed an infant with ankyloglossia.(16)

Ultrasound studies suggest that the mechanism of poor feeding in tongue-tied infants is due to restricted tongue movement, such that it may cause pain and/or trauma of the nipple, poor milk removal and unsustainable attachment to the breast. Frenotomy appears to restore sucking movements more analogous to breastfeeding infants without tongue-tie.(19)

ANKYLOGLOSSIA AND BREASTFEEDING DIFFICULTIES

Several studies have examined the association of ankyloglossia with breastfeeding difficulties. One study compared the rates of ankyloglossia in infants attending outpatient clinics for breastfeeding difficulties with the general population of normal term newborns, and reported a higher incidence of ankyloglossia in infants with breastfeeding difficulties (12.8% versus 3.2%).(8)

Another study recruited a cohort of 201 newborns with ankyloglossia and reported a high incidence of feeding difficulties (44%), but did not find a relationship between tongue-tie length and breastfeeding difficulties. This study also demonstrated that 56% of infants with tongue-tie can still feed adequately.(6)

One prospective trial showed a higher incidence of latching difficulties (19% versus 0%) and breastfeeding difficulties (25% versus 3%) in a group of 36 neonates with ankyloglossia compared with a control group of neonates with no ankyloglossia. Thirty (83%) of the 36 infants with ankyloglossia were successfully breastfed during the study period, compared with 33 (92%) of the 36 control infants (P=0.29). The duration of breastfeeding was similar in both groups.(9) This study also found no significant difference between ankyloglossia grades (moderate versus mild) or frenulum thickness in infants experiencing breastfeeding difficulties.(9)

MANAGING ANKYLOGLOSSIA

Management of tongue-tie is usually conservative, requiring no intervention beyond parental education, lactation support and reassurance. In cases of ankyloglossia and significant breastfeeding difficulties, there is some evidence that frenotomy can improve feeding. It remains controversial which tongue-ties need to be surgically released and which can be left to observation.

Several studies, including recent randomized controlled studies, have been conducted to evaluate the effectiveness of frenotomy in the setting of ankyloglossia with breastfeeding difficulties (Table 1).(6,8,20-27)

The limitations of these randomized controlled trials and prospective trials are substantial, and include the following:

- Variability and poor definition for diagnosing ankyloglossia, leading to unclear inclusion criteria;
- Small trial size;
- Most studies involved near-total crossover of the control group to the frenotomy arm of the study, precluding a fair assessment of outcomes and making it difficult to interpret findings;
- The objective outcome measurements were often limited and based on the observation of one feed;
- No reports of demographic information were included (ie, first-time versus experienced mothers with later birth order infants);
- Poorly defined outcomes (eg, 'feeding improvement') in some cases.

Blinding of observers and mothers in such studies is very difficult to achieve. In one study, 100% of supposedly masked experienced mothers correctly identified division of tongue-tie in their infant.(22) Careful consideration must also be given to the ability of a new mother to respond objectively about improved breastfeeding when her infant has just undergone a procedure to which she consented.

Furthermore, there is a surprising paucity of literature describing the 'normal' breastfeeding learning curve for mother and infant. This lack, along with the fact that a control group was seldom preserved during trials, makes it difficult to determine whether breastfeeding difficulties would have improved with time and conservative management (ie, natural history).

In addition to the studies described, several other prospective cohort studies have shown an association of ankyloglossia with breastfeeding difficulties(28) as well as the benefit of frenotomy in infants with ankyloglossia who present with breastfeeding difficulties.(10,29-32) However, these studies also share some of the limitations cited above.

Therefore, while several randomized trials and some cohort and cross-sectional prospective studies have shown some effectiveness for frenotomy in newborns who are having difficulties with breastfeeding due to ankyloglossia, they all have significant limitations.

FRENOTOMY PROCEDURE

If a tongue-tie release is deemed necessary, a referral to an otolaryngologist or a physician with experience performing this procedure should be made. Appropriate analgesia should be provided for the procedure. Unfortunately, there is also a paucity of literature regarding effective analgesia for frenotomy. Case reports have cited the use of acetaminophen, lidocaine and sucrose for analgesia, but none of these have been studied. Benzocaine was studied in a randomized controlled trial and was shown to be ineffective compared with placebo.(33) Release of the tongue-tie appears to be a minor procedure, but may cause complications such as bleeding, infection

TABLE 1
Studies examining the effectiveness of frenotomy in infants with ankyloglossia and breastfeeding (BF) difficulties

Authors (reference), year	Study type	Study population	Definition of ankyloglossia	Methods	Outcome measures	Results
Hogan et al (6), 2005	Randomized controlled trial	57 infants with ankyloglossia and feeding difficulties (breast or bottle) Mean age at randomization: 20 days	Percentage of tongue-tie gauged by eye, ranging from 100% (ie, to the tip) to 25%	Randomly assigned to frenotomy or control (advice) If no improvement noted in conservative group after 48 h, frenotomy offered	Telephone interview with mother at 24 h, weekly for four weeks and at four months Latching problems Sore nipples Continuous feeds Top-up feeds	27/28 infants randomly assigned to the frenotomy group improved their symptoms after procedure compared with 1/29 in control group 28/29 infants in control group underwent frenotomy Mothers of 27/28 of these infants had improvement in symptoms
Dollberg et al (20), 2006	Randomized, masked prospective study	25 infants (one to 21 days of age) with ankyloglossia and mothers with sore nipples	"The inability of the infant to protrude the tip of the tongue over the lower gum line while the tip was tied to the floor of the mouth by a tight cord of frenulum, and the tongue became heart-shaped when lifted up"	Randomized to one of two sequences: i) Frenotomy, BF, sham, BF (14 infants), or ii) Sham, BF, frenotomy, BF (11 infants)	After the first procedure (frenotomy or sham), a standardized LATCH score (21) was assessed and a pain scale assessment (1 to 10) was obtained from the mother	Maternal pain score decreased from 7.1 to 5.3 (CI overlap) after frenotomy and insignificant increase in LATCH score
Berry et al (22), 2012	Randomized, double-blinded controlled trial	60 breastfed infants (mean age 32 days) with BF problem (defined as difficulty with latch, nipple pain/trauma or inefficient feeding) and ankyloglossia	"Tongue-tie was present"	Randomized to immediate frenotomy (30 infants) or nonfrenotomy (30 infants)	Preprocedure: LATCH scoring and Infant Breastfeeding Assessment Tool (IBFAT) (23) and maternal pain score (1 to 10) during the sample feed Postprocedure: (First feed reportedly blinded): Objective assessment as above Subjective maternal assessment and pain score (1 to 10) Telephone call at one day and three months for subjective change in feeding, complications and BF rates	21/27 (78%) of mothers in frenotomy group reported subjective improvement following procedure compared with 14/30 (47%) of infants in control group Objective observer reported no statistical significance in feeding (50% improved in frenotomy group versus 40% in control group) 30 infants in nonfrenotomy group later underwent procedure At three months after frenotomy, 56% (33/59) reported full resolution, and 8% (5/59) no improvement BF rate at three-month follow-up was 51%
Emond et al (24), 2014	Randomized controlled trial	107 term infants (median age 11 days) with a mild or moderate degree of tongue-tie and difficulties with BF (defined by LATCH score ≤8)	Hazelbaker Assessment Tool for Lingual Frenulum Function Score (HATLFF) 6–12	Randomized to immediate frenotomy or standard care	Primary outcome: LATCH score at five days Secondary outcomes: LATCH score at eight weeks and the Infant Breast Feeding Assessment Tool score at five days and eight weeks, Breastfeeding Self-Efficacy Score – Short Form (BSES-SF) (25) and pain scale (1 to 10) at five days and eight weeks and infant weight at eight weeks	At five days, HATLFF score had increased in frenotomy group, but no difference in IBFAT, LATCH score or BSES-SF and pain 35/53 of control group offered frenotomy after five days No difference in any BF assessments or infant weight at eight weeks between groups BF rates of 80% in both groups at eight weeks
Ballard et al (8), 2002	Cohort study	Recruited 127 (of 3036 examined term infants inpatient and outpatient) with ankyloglossia	HATLFF function score >11/14 in the presence of an appearance score <8/10	123 infants underwent frenotomy (four mothers declined the procedure)	Latch not measured quantitatively; subjective by evaluators and mother Maternal nipple pain score (1 to 10)	Ankyloglossia accounted for 35/273 (12.8%) of BF problems seen at outpatient clinic Mean HATLFF scores similar for presenting features of poor latch and nipple pain Maternal nipple pain decreased postprocedure (6.9 to 1.2)

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TABLE 1 – CONTINUED
Studies examining the effectiveness of frenotomy in infants with ankyloglossia and breastfeeding (BF) difficulties

Authors (reference), year	Study type	Study population	Definition of ankyloglossia	Methods	Outcome measures	Results
Buryk et al (26), 2011	Randomized, single-blinded controlled trial	Infants with difficulty with BF and significant ankyloglossia (randomized at mean age of six days)	HATLFF function score >11/14 with failing lactation management or an appearance score <8/10	Randomized to frenotomy (30 infants) or a sham procedure (28 infants)	BF assessed pre- and postintervention Short-Form McGill Pain Questionnaire (SF-MPQ) (27) nipple pain scale and IBFAT and at two weeks and regular follow-ups Secondary outcome of effect of frenotomy on length of BF	Both groups had statistically significant decreased nipple pain scores after the intervention IBFAT score not significantly different between frenotomy and control groups postintervention 27/28 infants in sham group underwent frenotomy at or before time of two-week follow-up No difference in duration of BF Maternal subjective report of improved latch postfrenotomy

or injury to Wharton's duct. From the limited literature, the incidence of minor complications appears to be rare.(22)

A simple incision or 'snipping' of a tongue-tie (frenotomy) is the most common procedure performed for partial ankyloglossia. There is a risk that postoperative scarring may limit tongue movement even further, necessitating reoperation. Excision with lengthening of the ventral surface of the tongue, or a frenuloplasty release, are more complicated procedures. Both entail less postoperative scarring but carry the inherent risks associated with general anesthesia.(33,34)

Specialized private clinics are now performing frenotomy by laser ablation, but available data regarding the safety or efficacy of this procedure are limited.

CONCLUSION

Ankyloglossia is relatively common in the newborn population. Most of the time, ankyloglossia is an anatomical finding without significant consequences for infants affected by this condition. Current evidence appears to show that most newborns with this condition are still able to breastfeed successfully.

Based on available evidence, frenotomy cannot be recommended for all infants with ankyloglossia. There is no absolute relationship between ankyloglossia and breastfeeding difficulties. If an association between significant tongue-tie and major breastfeeding problems is identified and surgical intervention is deemed to be necessary, frenotomy should be performed by a clinician experienced with the procedure, using appropriate analgesia.

Consultation with a health care professional who has expertise in breastfeeding is recommended before referring a child for frenotomy.

RECOMMENDATIONS

Clear criteria are needed for the diagnosis of ankyloglossia, along with specific attention to characteristics of infants for whom a frenotomy would be of value to improve feeding. Identifying the specific characteristics of ankyloglossia that may guide the clinician in determining which infants are more likely to benefit from frenotomy is crucial for prognosis. Also, with respect to study design, future studies should include larger sample sizes and avoid crossover among study groups.

It is important to rule out other oral anomalies that may be causing breastfeeding difficulties. A thorough intraoral examination, including inspection of the tongue and its function, should be performed in newborns, particularly when there are feeding difficulties. The mother should then be interviewed regarding breastfeeding (latch, nipple pain, discomfort) and the feeding observed. If difficulties are identified, referral to a health care provider with experience in breastfeeding support should be considered.

REFERENCES

1. Messner AH, Lalakea ML. Ankyloglossia: Controversies in management. Int J Pediatr Otorhinolaryngol 2000;54(2-3):123-31.
2. Wright JE. Tongue-tie. J Paediatr Child Health 1995;31(4):276-8.
3. Griffiths DM. Do tongue ties affect breastfeeding? J Hum Lact 2004;20(4):409-14.
4. Kupietzky A and Botzer E. Ankyloglossia in the infant and young child: Clinical suggestions for diagnosis and management. Pediatr Dent 2005;27(1):40-6.
5. Rowan-Legg A; Canadian Paediatric Society, Community Paediatrics Committee. Ankyloglossia and breastfeeding. Paediatr Child Health 2011;16(4):222.
6. 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.
7. Ricke LA, Baker NJ, Madlon-Kay DJ, DeFor TA. Newborn tongue-tie: Prevalence and effect on breast-feeding. J Am Board Fam Pract 2005;18(1):1-7.
8. Ballard JL, Auer CE, Khoury JC. Ankyloglossia: Assessment, incidence, and effect of frenuloplasty on the breastfeeding dyad. Pediatrics 2002;110(5):e63.
9. Messner AH, Lalakea ML, Aby J, Macmahon J, Bair E. Ankyloglossia: Incidence and associated feeding difficulties. Arch Otolaryngol Head Neck Surg 2000;126(1):36-9.
10. 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.
11. Hazelbaker AK. The Assessment Tool for Lingual Frenulum Function (ATLFF): Use in a lactation consultant's private practice. Pasadena: Pacific Oaks College, 1993.
12. Amir LH, James JP, Donath SM. Reliability of the Hazelbaker assessment tool for lingual frenulum function. Int Breastfeed J 2006;1(1):3-8.
13. Segal LM, Stephenson R, Dawes M, Feldman P. Prevalence, diagnosis, and treatment of ankyloglossia: Methodologic review. Can Fam Physician 2007;53(6):1027-33.
14. 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.
15. Berg KL. Tongue-tie (ankyloglossia) and breastfeeding: A review. J Hum Lact 1990;6(3):109-12.
16. Geddes DT, Kent JC, McClellan HL, Garbin CP, Chadwick LM, Hartmann PE. Sucking characteristics of successfully breastfeeding infants with ankyloglossia: A case series. Acta Paediatr 2010;99(2):301-3.

17. Garbin CP, Sakalidis VS, Chadwick LM, Whan E, Hartmann PE, Geddes DT. Evidence of improved milk intake after frenotomy: A case report. Pediatrics 2013;132(5):e1413-17.
18. Wight NE. Management of common breastfeeding issues. Pediatr Clin North Am 2001;48(2):321-44.
19. Geddes DT, Langton DB, Gollow I, Jacobs LA, Hartmann PE, Simmer K. Frenulotomy for breastfeeding infants with ankyloglossia: Effect on milk removal and sucking mechanism as imaged by ultrasound. Pediatrics 2008;122(1):e188-94.
20. Dollberg S, Botzer E, Grunis E, Mimouni FB. Immediate nipple pain relief after frenotomy in breast-fed infants with ankyloglossia: A randomized, prospective study. J Pediatr Surg 2006;41(9):1598-600.
21. Jensen D, Wallace S, Kelsay P. LATCH: A breastfeeding charting system and documentation tool. J Obstet Gynecol Neonatal Nurs 1994;23(1):27-32.
22. 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.
23. Matthews MK. Developing an instrument to assess infant breastfeeding behaviour in the early neonatal period. Midwifery 1988;4(4):154-65.
24. 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.
25. Dennis CL. The breastfeeding self-efficacy scale: Psychometric assessment of the short form. J Obstet Gynecol Neonatal Nurs 2003;32(6):734-44.
26. Buryk M, Bloom D, Shope T. Efficacy of neonatal release of ankyloglossia: A randomized trial. Pediatrics 2011;128(2):280-8.
27. Melzack R. The short-form McGill Pain Questionnaire. Pain 1987;30(2):191-7.
28. 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.
29. Post E, Daamen J, Balemans W. Snipping of a "tongue tie" in neonates with ankyloglossia and breastfeeding problems: Outcomes and complications. Arch Dis Child 2012:97(Suppl 2):A486.
30. Kumar M, Kalke E. Tongue-tie, breastfeeding difficulties and the role of frenotomy. Acta Paediatr 2012;101(7):687-9.
31. Sethi N, Smith D, Kortequee S, Ward VM, Clarke S. Benefits of frenulotomy in infants with ankyloglossia. Int J Pediatr Otorhinolaryngol 2013;77(5):762-5.
32. Dollberg S, Marom R, Botzer E. Lingual frenotomy for breastfeeding difficulties: A prospective follow-up study. Breastfeed Med 2014;9(6):286-9.
33. Ovental A, Marom R, Botzer E, Batscha N, Dollberg S. Using topical benzocaine before lingual frenotomy did not reduce crying and should be discouraged. Acta Pediatr 2014;103(7):780-2.
34. Catlin FI. Tongue-tie. Arch Otolaryngol 1971;94(6):548-57.

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