

Tongue Tie and Frenotomy in the Breastfeeding Newborn

Isabella Knox, MD, EdM*

Author Disclosure
Dr Knox has disclosed no financial relationships relevant to this article. This commentary does not contain a discussion of an unapproved/investigative use of a commercial product/device.

Abstract

Tongue tie or ankyloglossia has been the subject of much controversy. As defined in this review, tongue tie occurs when a common minor embryologic tissue remnant—persistence of midline sublingual tissue that usually undergoes apoptosis during embryonic development—causes restriction of normal tongue movement. Effective breastfeeding requires newborns to fine-tune their tongue movements to adapt to their mothers' particular nipple and breast anatomy and physiology. In the presence of tongue tie, two categories of signs/symptoms arise: those related to nipple trauma and those related to ineffective breast emptying and low infant intake. Untreated tongue tie can lead to untimely weaning and its attendant health risks. Frenotomy is a safe and effective procedure to release tongue tie and improve tongue function and breastfeeding outcomes.

Objectives After completing this article, readers should be able to:

1. Recognize the existence of tongue tie as a condition that can potentially cause clinical dysfunction.
2. Understand the pathophysiologic mechanisms of clinical signs and symptoms of tongue tie in a breastfeeding dyad.
3. Determine when to refer a breastfeeding infant for frenotomy.

Introduction

Tongue tie, also known as ankyloglossia, occurs when a persistent lingual frenum restricts tongue movements and interferes with tongue function. True ankyloglossia is classified as a minor congenital anomaly. (1) Controversy has existed for centuries about if and when sublingual frenum tissue is the actual cause of clinical dysfunction. Over the past 3 decades, advances have been made in defining the embryology, pathophysiology, clinical features, and treatment of tongue tie. It is now clear that in some cases sublingual frenum tissue causes tongue tie because the tissue is restrictive and creates difficulties with breastfeeding and, later in life, other activities that require tongue mobility. The field is still hampered by the lack of a uniform definition, diagnostic criteria, treatment indications, and outcomes research. This review summarizes current knowledge about tongue tie in 2010.

History

Tongue tie is mentioned in the Bible and other written documents that precede the modern medical literature. (2)(3) For many centuries, it was believed that the sublingual frenum could impair an infant's ability to extract milk from the breast and, hence, to survive. Frenotomy was widely practiced by midwives and physicians until the mid-20th century, coinciding with the period in which few infants in the United States were breastfed. (4) Tongue tie rarely causes major dysfunction in bottlefeeding infants, and it was not until the early 1990s, after breastfeeding rates had risen significantly, that tongue tie and frenotomy reappeared in the medical literature. Tongue tie is now confirmed as a relatively common and remediable clinical entity, (5)(6)(7)(8) although many important clinical questions remain. In 2010, all neonatologists, general pediatricians, and family physicians should understand this entity and ensure that resources are available to their patients for diagnosis and remediation.

*Department of Pediatrics, Division of Neonatology, University of Washington, Seattle, Wash.

Embryology

Between the fourth and seventh weeks of development, the oral cavity of the embryo develops from a smooth-walled tube to a more mature structure that has complex anatomy, including a mobile, muscular tongue that protrudes freely from the floor of the mouth (Fig. 1). The tongue develops from bilateral tissue buds that grow from the inner surface of the oral tube, fuse posteriorly-to-anteriorly, and separate from the floor of the mouth. As with many processes in embryologic development, the separation occurs by a combination of tissue growth and programmed morphologic cell death (apoptosis). Similar to interdigital synchia seen in soft-tissue syndactyly, (9) the persistent lingual frenum is likely a remnant of incomplete apoptosis. (10)(11) This anomaly most commonly occurs in isolation. Its association with cleft palate has been

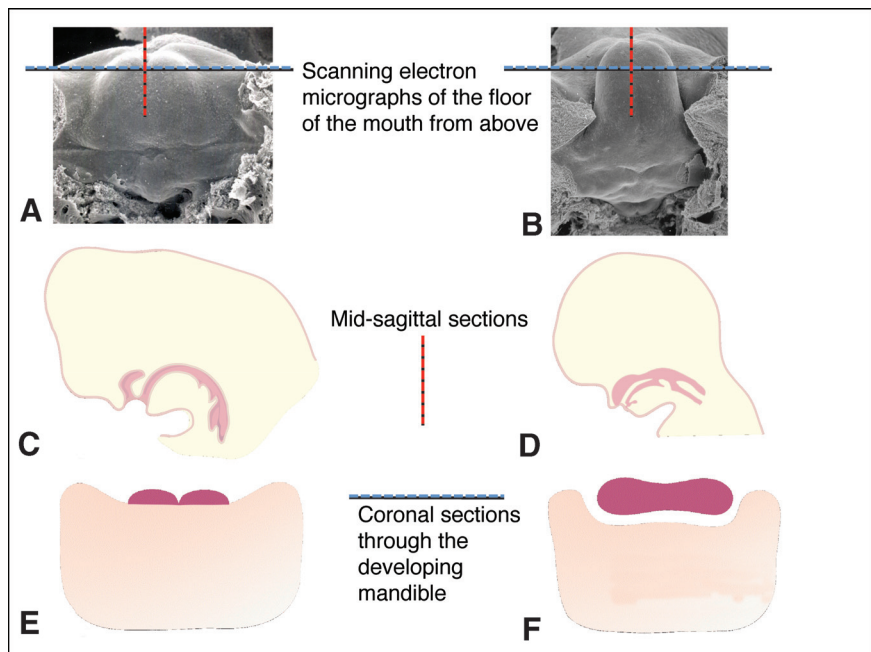


Figure 1. Embryology of tongue development. A. and B. Scanning electron micrographs of mouse embryo at days 10 and 12, respectively, approximately equivalent to human weeks 5 and 7. Courtesy of K. K. Sulik, all rights reserved. C. and D. Corresponding mid-sagittal anatomy. Drawn by Kelly Ledbetter, © 2010, University of Washington. E. and F. Corresponding coronal sections through the anterior tongue.

Terminology

Frenum (or frenulum):	A membranous fold of skin or mucous membrane that supports or restricts the movement of a part of organ.
Persistent lingual frenum:	Presence of a frenum between the underside of the tongue and the floor of the mouth; this does not necessarily cause clinical dysfunction.
Tongue tie:	Restriction of tongue movement or function by a persistent lingual frenum. Note that some authors use “tongue tie” to refer to the presence of <i>any</i> sublingual tissue. In this article, a clear distinction is made between frenum tissue, which may be present and in no way interfere with tongue function, and tongue tie, in which the frenum is restrictive.
Ankyloglossia:	Synonym for tongue tie.
Frenotomy (or frenulotomy):	Surgical procedure in which the frenum is incised.
Frenectomy (or frenulectomy):	Surgical procedure in which frenum tissue is excised.

well characterized, including identification of an underlying *TBOX* gene abnormality in some kindreds. (12) Other anomalies have been associated with tongue tie, most often involving craniofacial structures. Maternal cocaine use has been documented as a risk factor for tongue tie. (13)

Epidemiology

The incidence of tongue tie is difficult to ascertain because of the lack of a uniform assessment strategy. Studies that rely on visual inspection alone report a rate of persistent lingual tissue of 3% to 10%. (5)(8) Several studies have attempted to assess the risk that the presence of this tissue poses for breastfeeding

difficulties (ie, tongue tie) but are complicated by the large numbers needed and the many factors involved in breastfeeding success. The risk appears to be significant, in the range of 25% to 60%. (14) In most studies, a male:female ratio of about 2:1 has been reported, although Knox and O'Callahan recently reported the first large series of posterior tongue ties and found a male:female ratio of 1:1. (15)

Pathophysiology

Complex tongue movements are critical for successful breastfeeding. Each mother and baby have unique anatomic features, and the infant must adapt his or her sucking behavior accordingly. To extract milk successfully, the tongue must undertake several actions (Fig. 2 and Video 1 in [data supplement](#)):

1. Protrude over the alveolar ridge both to inhibit the bite reflex (16) and to contribute to an airtight seal on the areola that allows creation of an intraoral vacuum.
2. Manipulate the nipple and areolar tissue into the

proper relationship with the hard and soft palates, the tongue itself, and the swallowing/breathing apparatus.

3. Produce the intraoral vacuum that results in milk flow from the breast. (17)

Restriction of the tongue's ability to move freely leads to suboptimal nursing mechanics. The nipple's spatial relationship to the infant's mouth structures and the specific pattern of tongue movements are critical both for effective and efficient milk removal and for protection of the delicate nipple tissue from trauma. The downstream effects of tongue tie are related either to ineffective breast emptying or to nipple trauma. All of the common signs and symptoms of tongue tie can be explained by the effects of abnormal sucking mechanics (Table).

Diagnostic Features

In addition to the clinical signs and symptoms listed in Table, the diagnosis of tongue tie is based on anatomic and functional abnormalities found on physical examination. The physical examination may reveal either a thin anterior membrane or a thick posterior fibrous strand in the midline between the underside of the tongue and the floor of the mouth (Fig. 3). The superior attachment of the frenum to the tongue can occur in various locations between the tip and the junction with the floor of the mouth. On occasion, no abnormality is visible. Instead, a tight midline filament, like a taut fishing line in the soft tissue where the tongue joins the floor of the mouth, is palpable. In general, anterior frena are thin and transparent, merely consisting of a reflection of mucosal tissue, while posterior frena are thicker, white, and fibrous-looking. Occasionally, blood vessels or muscle fibers may be present in the frenum.

Assessment of newborn tongue function is challenging, particularly during nursing, when most structures are not visible. Because maternal characteristics also contribute to the structural mechanics of the infant's task (eg, the length, firmness, and stretchability of the nipple and

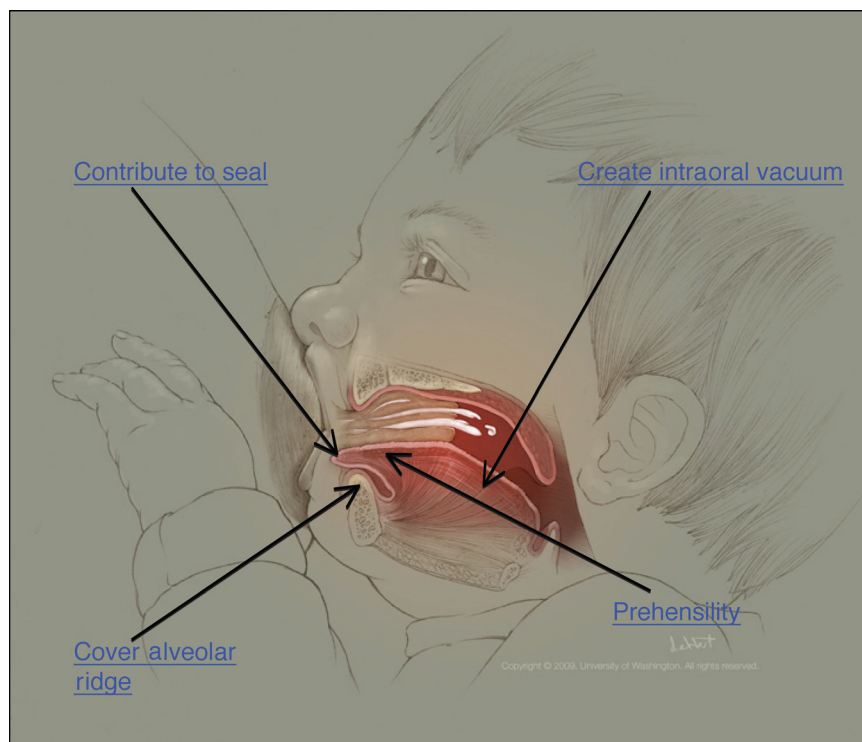


Figure 2. During nursing, the infant's tongue must: 1) protrude over the alveolar ridge to inhibit the bite reflex, 2) assist the flanged lips in maintaining an airtight seal on the areola, 3) with its prehensile function, manipulate the nipple into the proper position (note the depth of the nipple tip and its proximity to the hard-soft palate junction), and 4) via distal-to-proximal muscular contractions that end with the lowering of the base of the tongue, produce an intraoral vacuum, resulting in milk flow.

Table. Signs and Symptoms of Tongue Tie in the Breastfeeding Dyad

Infant Signs/Symptoms	Mechanism
Poor latch	Inadequate seal, inability to manipulate nipple/areola into position
Clicking sound while nursing	Intermittent loss of seal
Gradual sliding off breast	Inability to maintain hold on nipple/areola
"Chewing"	Failure to inhibit bite reflex; compensatory use of jaw muscles to maintain nipple position
Ineffective milk transfer	Failure of breast emptying
Poor weight gain or weight loss	Failure of milk transfer
Hypernatremic dehydration (18)	Unrecognized severe failure of milk transfer
Fussiness and arching away from breast	Infant response to frustration
Falling asleep at breast	Infant exhaustion
Maternal Signs/Symptoms	Mechanism
Nipple trauma: pain, blister, crack, bleeding, scab	Localized ischemia from sustained point compression; tissue destruction from repetitive crush or roll, excessive vacuum (19)
Painful breasts	Engorgement from failure to empty during primary lactogenesis; infection
Low milk supply	Failure of secondary lactogenesis from failure to empty
Plugged ducts	Stasis from failure to empty
Mastitis	Failure to empty with bacterial ingress from nipple tissue barrier break
Frustration, disappointment, discouragement about breastfeeding	Pain, infant frustration, sense of failure because of inability to nourish infant
Untimely weaning	Lack of availability of tongue tie diagnosis and remediation

areolar tissue), it is difficult to predict from examination of the infant whether or not he or she will be able to nurse effectively with his or her particular mother. Hazelbaker (20) developed a scoring system, the Assessment Tool for Lingual Frenulum Function, that includes assessment of structural and functional aspects of the infant's tongue. Use of this tool is not widespread, possibly because of its complexity and the large number of items to be scored. In this

author's opinion, as well as that of others, including the United Kingdom's National Institute for Health and Clinical Excellence, (21)(22) if a thin anterior frenum is found in association with clinical signs and symptoms, frenotomy should be considered because the procedure is simple and safe and may solve the problem. Posterior frena are more likely than thin anterior ones to be associated with breastfeeding difficulties (4) and have received more attention

recently. In addition, anecdotal reports suggest that thick posterior frenum tissue may remain after anterior membrane frenotomy (personal communication, E. Coryllos, C. O'Callahan, J. Murphy, 2010), so persistent tongue tie should be considered if signs and symptoms do not resolve. Submental ultrasonography allows viewing of nipple-tongue relationships and tongue movement. Although this tool so far has been used only for research, it may, in the future, be helpful in diagnosis, particularly when clinical findings are equivocal.

Treatment

When tongue tie is associated with breastfeeding difficulties, frenotomy

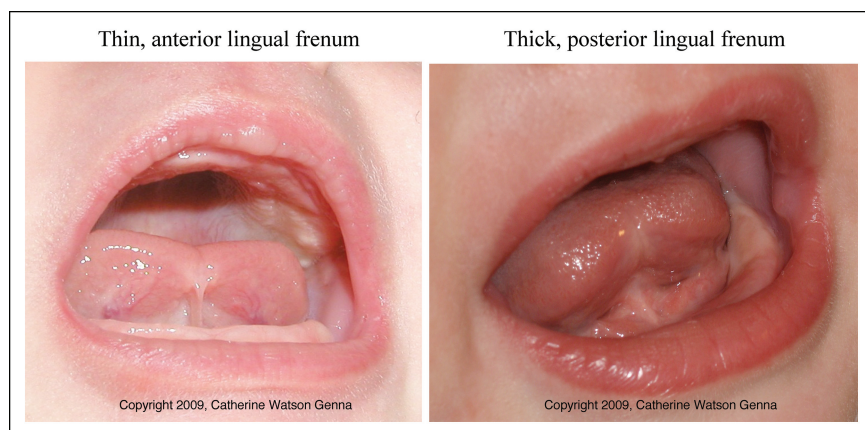


Figure 3. The two ends of the ankyloglossia spectrum. On the left is a classic thin anterior frenum. Note the notch in the tongue tip and the limited height of the tongue lift. On the right is a thick posterior frenum, visible as a white line in the posterior floor of the mouth. On palpation, this tissue is taut and firm. Pictures courtesy of Catherine Watson Genna, used with permission, all rights reserved.

is an effective treatment. Frenotomy for thin anterior tongue tie is a safe and simple nursery or office procedure that uses readily available equipment (see Video 2 in the [data supplement](#)). (23) Frenotomy for thick posterior tongue tie currently is performed either as an office procedure or in the operating room, depending on the experience and training of the operator. Specialized training is required to perform frenotomy for posterior tongue tie safely.

Because few physicians are knowledgeable about tongue tie and even fewer perform frenotomies, only the most persistent and knowledgeable families can take advantage of this treatment and, thus, maintain breastfeeding. Historically, severe bleeding and infection were reported as complications of frenotomy. (24)(25) There is also a potential for damage to the sublingual salivary ducts. However, all reported series of newborn and infant frenotomy in the modern medical literature, including those cited in this article's reference list, (5)(6)(7)(8) state that no complications occurred related to the procedure.

After frenotomy, the infant should breastfeed as soon as hemostasis has been assured, usually after one blot with a gauze pad, as seen in Video 2. Tongue movements during nursing provide range of motion exercise and help to prevent re-adhesion. Skilled lactation support must continue until the infant establishes functional motor patterns and injured nipples heal. Some infants have difficulty developing normal tongue movements; feeding specialists can provide detailed assessment and targeted therapy.

Outcomes of Treated and Untreated Tongue Tie

The effectiveness of frenotomy for tongue tie-associated breastfeeding difficulties has been documented in observational (5) and randomized (6)(8) studies, including one that was well-blinded. (6) Geddes and associates (7) used ultrasonography to document improvement in tongue mechanics following fre-

notomy, along with improved milk transfer, decreased nipple pain, and better latch (Fig. 4). Studies of neonatal and infant frenotomy have examined only its short-term effects on breastfeeding parameters. It is unknown whether frenotomy in infancy alters the natural history of untreated tongue tie. Dollberg and colleagues (26) examined speech at age 8 years in infants treated with neonatal frenotomy. Their speech was not different from children who had not had tongue tie. A comparison group with untreated tongue tie had significant misarticulations.

Untreated tongue tie is associated with nursing difficulties that may result in early weaning, depriving families and infants of the well-documented short- and long-

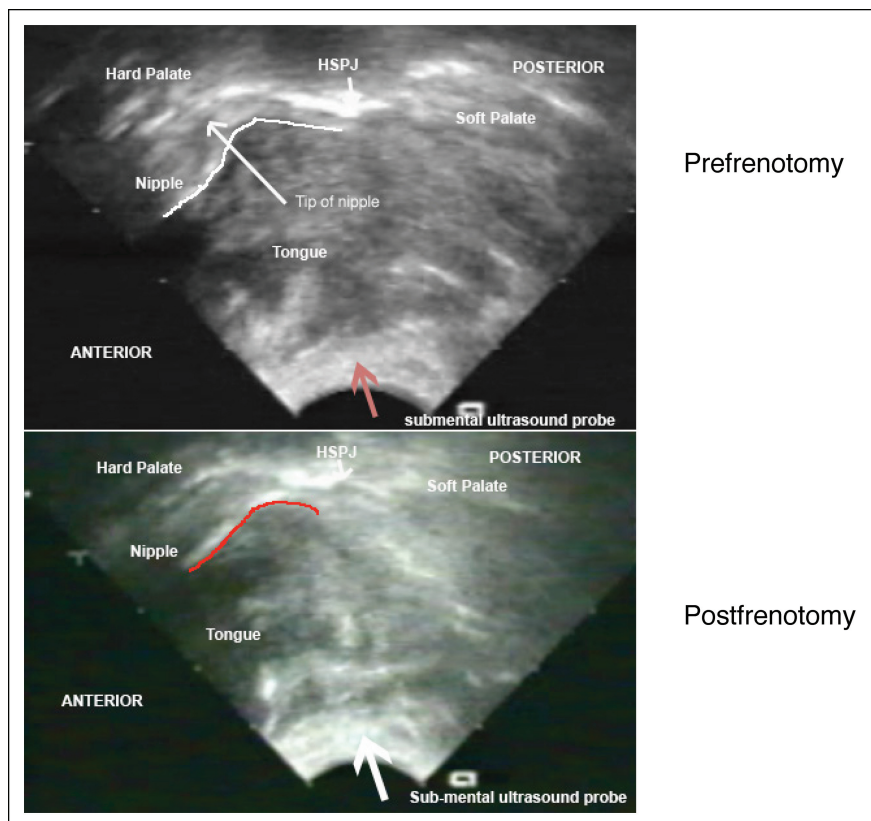


Figure 4. Submental ultrasonographic images obtained pre- and postfrenotomy. The orientation is the same as in Video 1. Both images were obtained in the “tongue up” portion of the suck cycle. In this patient, the primary findings were an increased distance from the tip of the nipple to the hard-soft palate junction (HSPJ) and compression of the tip of the nipple by the back of the tongue. The second image was obtained 1 week after the procedure. Notable are the shortened (and normal) distance between the nipple and the HSPJ and the smoother contour of the back of the tongue, which no longer is deforming the nipple. For more information about ultrasonographic assessment in tongue tie, see Geddes and associates. (7)(17) Images provided by Donna T. Geddes, PhD, all rights reserved.

term benefits of breastfeeding. There are anecdotal reports of spontaneous rupture of the lingual frenum. (27) Clinical problems associated with tongue tie in older children and adults have been documented in case reports and series. (28) Problems include difficulties with speech, tooth alignment requiring orthodontia, oral hygiene (licking lips and removing debris, especially from posterior teeth), and kissing. Some, although not all, of these problems improved following frenectomy. No controlled studies are available.

Summary and Conclusions

When the lingual frenum does not apoptose properly during embryologic development, the residual tissue can, in some cases, interfere with tongue function and cause a variety of breastfeeding problems that may result in early weaning and its attendant risks. Persistence of this tissue beyond infancy can interfere with full tongue function in the older child and adult. Every mother-infant dyad with breastfeeding problems, particularly the maternal symptom of nipple pain and the infant sign of failure to transfer milk despite vigorous attempts to latch and suckle, should be evaluated for tongue tie and treated if it is present. Treatment ideally should occur within 24 hours of diagnosis because of the fragility of lactation physiology in the neonatal period.

Frenotomy is an effective treatment and, for thin anterior frena, is a simple and safe procedure. The procedure must be followed by skilled lactation and oral skill development support until breastfeeding is re-established. Thick posterior tongue ties are more difficult to diagnose and treat.

Frenotomy is not as widely available as is needed. Because facilitating the full breastfeeding potential of all mother-baby dyads is critical to individual and public health, more practitioners need to be trained to assess tongue tie and perform frenotomy. Neonatologists, with specific procedural skills and commitment to breastfeeding, can lead the way as advocates and teachers. More research is needed, particularly on posterior tongue tie, to facilitate and standardize diagnosis and treatment.

Follow-up studies are needed to examine the long-term outcome of frenotomy in infancy.

References

1. Ankyloglossia. MIM 106280. *Online Mendelian Inheritance in Man*. 2009
2. Dunn PM. Bridled babies: a history of tongue tie. *Proc Bristol Medico-Historical Soc*. 1995–1999;3:15–23
3. Obladen M. Much ado about nothing: two millennia of controversy on tongue-tie. *Neonatology*. 2010;97:83–89
4. Centers for Disease Control and Prevention. *Breastfeeding Among US Children Born 1999–2006. CDC National Immunization Survey*. 2010. Accessed June 2010 at: http://www.cdc.gov/breastfeeding/data/NIS_data/index.htm
5. Ballard J, Auer C, Khoury J. Ankyloglossia: assessment, incidence and effect of frenuloplasty on the breastfeeding dyad. *Pediatrics*. 2002;110:e63
6. Dollberg S, Botzerb E, Gunisa E, Mimounia FB. Immediate nipple pain relief after frenotomy in breast-fed infants with ankyloglossia: a randomized, prospective study. *J Pediatr Surg*. 2006;41:1598–1600
7. 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:e188–e194
8. Hogan M, Westcott C, Griffiths M. Randomized, controlled trial of division of tongue-tie in infants with feeding problems. *J Paediatr Child Health*. 2005;41:246–250
9. Chen Y, Zhao X. Shaping limbs by apoptosis. *J Exp Zool*. 1998;282:691–702
10. 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;24:9736–9743
11. Karahan S, Cinar Kul B. Ankyloglossia in dogs: a morphological and immunohistochemical study. *Anat Histol Embryol*. 2009;38:118–121
12. Braybrook C, Lisgo S, Doudney K, et al. Craniofacial expression of human and murine TBX22 correlates with the cleft palate and ankyloglossia phenotype observed in CPX patients. *Hum Mol Genet*. 2002;11:2793–2804
13. Harris EF, Friend GW, Tolley EA. Enhanced prevalence of ankyloglossia with maternal cocaine abuse. *Cleft Palate Craniofacial J*. 1992;29:72–76
14. Segal LM, Staphenson R, Dawes M, Feldman P. Prevalence, diagnosis, and treatment of ankyloglossia. *Can Fam Physician*. 2007;53:1027–1033
15. Knox I, O'Callahan C. Posterior tongue tie: new entity or on the ankyloglossia spectrum? Submitted to the Pediatric Academic Society Annual Meeting, Vancouver, May 2010
16. Coryllos E, Genna CW, Salloum AC. Congenital tongue-tie and its impact on breastfeeding. *Newsletter of the American Academy of Pediatrics Section on Breastfeeding*. 2004; Summer. Accessed June 2010 at: www.aap.org/breastfeeding/files/pdf/BBM-8-27%20Newsletter.pdf
17. Geddes DT, Kent JC, Mitoulas LR, Hartmann PE. Tongue movement and intra-oral vacuum in breastfeeding infants. *Early Hum Dev*. 2008;84:471–477
18. Livingstone VH, Willis CE, Abdel-Wareth LO, Thiessen P, Lockitch G. Neonatal hypernatremic dehydration associated with

American Board of Pediatrics Neonatal-Perinatal Medicine Content Specification

- Recognize common problems associated with breast milk production in the NICU, and their management.



breast-feeding malnutrition: a retrospective survey. *Can Med Assoc J.* 2000;162:647–652

19. McClellan H, Geddes D, Kent J, Garbin C, Mitoulas L, Hartmann P. Infants of mothers with persistent nipple pain exert strong sucking vacuums. *Acta Paediatr.* 2008;97:1205–1209

20. Hazelbaker AK. The Assessment Tool for Lingual Frenulum Function (ATLFF): use in a lactation consultant private practice [thesis]. Pasadena, Calif: Pacific Oaks College; 1993

21. Algar V. Should an infant who is breastfeeding poorly and has a tongue tie undergo a tongue tie division? *Arch Dis Child.* 2009; 94:911–912

22. National Institute for Health and Clinical Excellence. *Interventional Procedure Guidance 149. Division of Ankyloglossia (Tongue Tie) for Breastfeeding.* London, United Kingdom: NICE; 2005

23. Jain E. *Tongue-Tie: Impact on Breastfeeding: Complete Man-*

agement Including Frenotomy. Los Angeles, Calif: Geddes Productions; 2001. Accessed June 2010 at: www.geddesproduction.com

24. Butlin HT, Spencer WG. *Diseases of the Tongue.* New York, NY: Cassell & Co; 1900:30–32

25. Allbutt TC. *A System of Medicine.* London, United Kingdom: MacMillan & Co; 1907:321

26. Dollberg S, Manor Y, Makai E, Botzer E. Evaluation of speech intelligibility in children with tongue-tie [abstract.] Presented at Pediatric Academic Society Annual Meeting, Baltimore, May 2009

27. Smithells RW. Spontaneous rupture of tongue tie. *Br Med J.* 1959;1:236

28. Messner AH, Lalakea ML. Ankyloglossia: the adolescent and adult perspective. *Otolaryngol Head Neck Surg.* 2003;128: 746–752