Abstract
Ankyloglossia (‘tongue-tie’) is a relatively common congenital anomaly characterized by an abnormally short lingual frenum, 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 frenum). 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

Background
Ankyloglossia (‘tongue-tie’) is a congenital anomaly observed in newborns and children, and is characterized by an abnormally short lingual frenum. The tight frenum 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.4 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 frenum (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).1-9 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 frenum (Type III) or a submucosal frenum (a flat, broad mound) that restricts movement at the base of the tongue (Type IV).6 These definitions are seldom used in the literature and rarely in the clinical domain. Hazelbaker10 developed a descriptive assessment tool for lingual frenum function; however, it is complex, lengthy and has not been
valuated in a controlled manner.\textsuperscript{12} Criteria used to di-
gnose ankyloglossia show considerable variation, and
there is no accepted standard. The lack of a consistent
definition further fuels controversy regarding this condi-
tion and its clinical significance.

**Etiology**

The tongue is fused to the floor of the mouth in early de-
velopment. Cell death and resorption free the tongue,
with the frenulum left as the only remnant of initial at-
tachment. 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.\textsuperscript{2} This process oc-
curs 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.\textsuperscript{2}

There may be a genetic predisposition to ankylogloss-
na.\textsuperscript{13} This congenital anomaly typically occurs in iso-
lolation.

**Prevalence**

The reported prevalence of ankyloglossia in infants is
variable in the literature, reflecting the lack of a consis-
tent definition. Estimates range from 4.2% to 10.7% in
newborns.\textsuperscript{7,19}

**Pathophysiology**

The role of a short lingual frenum 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.\textsuperscript{14-17} To nurse suc-
cessfully, 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.\textsuperscript{18} It is clear that re-
striction of tongue movement must be extreme to in-
terfere with sucking and swallowing.\textsuperscript{14} It also appears
that some mothers have particular breast, nipple or milk
ejection characteristics that allow them to successfully
breastfeed an infant with ankyloglossia.\textsuperscript{18}

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 at-
tachment to the breast. Frenotomy appears to restore
sucking movements more analogous to breastfeeding
infants without tongue-tie.\textsuperscript{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 attend-
ing 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%).\textsuperscript{19}

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 be-
tween tongue-tie length and breastfeeding difficulties.
This study also demonstrated that 56% of infants with
tongue-tie can still feed adequately.\textsuperscript{8}

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 breastfeed-
ing was similar in both groups.\textsuperscript{19} This study also found
no significant difference between ankyloglossia grades
(moderate versus mild) or frenulum thickness in infants
experiencing breastfeeding difficulties.\textsuperscript{9}

**Managing ankyloglossia**

Management of tongue-tie is usually conservative, re-
quiring no intervention beyond parental education, lac-
tation support and reassurance. In cases of ankyloglos-
sia and significant breastfeeding difficulties, there is
some evidence that frenotomy can improve feeding. It
remains controversial which tongue-ties need to be sur-
gically released and which can be left to observation.

Several studies, including recent randomized controlled
studies, have been conducted to evaluate the effective-
ness of frenotomy in the setting of ankyloglossia with
breastfeeding difficulties (Table 1).\textsuperscript{18,19,20-22}
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. 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 as well as the benefit of frenotomy in infants with ankyloglossia who present with breastfeeding difficulties. 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. Release of the tongue-tie appears to be a minor procedure, but may cause complications such as bleeding, infection or injury to Wharton’s duct. From the limited literature, the incidence of minor complications appears to be rare.

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.

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.
<table>
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<tr>
<th>Authors (reference), year</th>
<th>Study type</th>
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<th>Definition of ankyloglossia</th>
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<tr>
<td>Hogan et al [6], 2005</td>
<td>Randomized controlled trial</td>
<td>57 infants with ankyloglossia and feeding difficulties (breast or bottle)  Mean age at randomization: 20 days</td>
<td>Percentage of tongue-tie gauged by eye, ranging from 100% (ie, to the tip) to 25%</td>
<td>Randomly assigned to frenotomy or control (advice)  If no improvement noted in conservative group after 48 h, frenotomy offered</td>
<td>Telephone interview with mother at 24 h, weekly for four weeks and at four months  Latching problems  Sore nipples  Continuous feeds  Top-up feeds</td>
<td>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</td>
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<td>Dollberg et al [20], 2006</td>
<td>Randomized, masked prospective study</td>
<td>25 infants (one to 21 days of age) with ankyloglossia and mothers with sore nipples</td>
<td>&quot;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&quot;</td>
<td>Randomized to one of two sequences: i) Frenotomy, BF, sham, BF (14 infants), or ii) Sham, BF, frenotomy, BF (11 infants)</td>
<td>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</td>
<td>Maternal pain score decreased from 7.1 to 5.3 (CI overlap) after frenotomy and insignificant increase in LATCH score</td>
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<td>Berry et al [22], 2012</td>
<td>Randomized, double-blind controlled trial</td>
<td>60 breastfed infants (mean age 32 days) with BF problem (defined as difficulty with latch, nipple pain/trauma or inefficient feeding) and ankyloglossia</td>
<td>&quot;Tongue-tie was present&quot;</td>
<td>Randomized to immediate frenotomy (30 infants) or nonfrenotomy (30 infants)</td>
<td>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</td>
<td>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)</td>
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<tr>
<td>Study</td>
<td>Design</td>
<td>Sample Description</td>
<td>Assessment Tool(s)</td>
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<td>Emond et al [24], 2014</td>
<td>Randomized controlled trial</td>
<td>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)</td>
<td>Hazelbaker Assessment Tool for Lingual Frenulum Function Score (HATLFF) 6–12</td>
<td>Randomized to immediate frenotomy or standard care</td>
<td>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</td>
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<td>Ballard et al [8], 2002</td>
<td>Cohort study</td>
<td>Recruited 127 (of 3036 examined) term infants inpatient and outpatient with ankyloglossia</td>
<td>HATLFF function score &gt;11/14 in the presence of an appearance score &lt;8/10</td>
<td>123 infants underwent frenotomy (four mothers declined the procedure)</td>
<td>Latch not measured quantitatively; subjective by evaluators and mother Maternal nipple pain score (1 to 10)</td>
<td>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)</td>
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Randomized, single-blind 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) 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

References

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