Prevalence, diagnosis, and treatment of ankyloglossia

Methodologic review

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ABSTRACT

OBJECTIVE To review the diagnostic criteria for, the prevalence of, and the effectiveness of frenotomy for treatment of ankyloglossia.

DATA SOURCES MEDLINE and CINAHL databases were searched for articles suitable for a methodologic review of studies on various aspects of ankyloglossia.

STUDY SELECTION Studies that presented data on patients and addressed ankyloglossia in relation to breastfeeding were selected. Case reports, case series, retrospective studies, prospective controlled studies, and randomized controlled trials were included in the analysis. Opinion pieces, literature reviews, studies without data on patients, studies that did not focus on breastfeeding, position statements, and surveys were excluded.

SYNTHESIS There is no well-validated clinical method for establishing a diagnosis of ankyloglossia. Five studies using different diagnostic criteria found a prevalence of ankyloglossia of between 4% and 10%. The results of 6 non-randomized studies and 1 randomized study assessing the effectiveness of frenotomy for improving nipple pain, sucking, latch, and continuation of breastfeeding all suggested frenotomy was beneficial. No serious adverse events were reported.

CONCLUSION Diagnostic criteria for ankyloglossia are needed to allow for comparative studies of treatment. Frenotomy is likely an effective treatment, but further randomized controlled trials are needed to confirm this. A reliable frenotomy decision rule is also needed.

RÉSUMÉ

OBJECTIF Faire le point sur les critères diagnostiques, la prévalence et l'efficacité de la frénotomie pour traiter l'ankyloglosse.

SOURCES DES DONNÉES On a repéré dans MEDLINE et CINAHL les articles pouvant servir à une revue méthodologique de divers aspects de l'ankyloglosse.

CHOIX DES ÉTUDES On a retenu les articles contenant des données sur les patients et qui portaient sur la relation entre l'ankyloglosse et l'allaitement. L'analyse incluait des rapports de cas, séries de cas, études rétrospectives, essais prospectifs avec témoins et essais contrôlés randomisés. On a exclu de l'analyse les articles d'opinion, les critiques des ouvrages spécialisés, les études ne comportant pas de données sur les patientes et celles qui ne portaient pas sur l'allaitement, les exposés de position et les sondages.

SYNTHÈSE Il n'existe pas de méthode clinique reconnue pour établir un diagnostic d'ankyloglosse. Cinq études qui utilisaient différents critères diagnostiques ont estimé que la prévalence de l'ankyloglosse se situait entre 4% et 10%. Les résultats de 6 études non randomisées et d'une étude randomisée ont tous démontré que la frénotomie a des effets favorables sur la douleur mamelonnaire, la succion, le verrouillage et la poursuite de l'allaitement. Aucun effet indésirable sérieux n'a été rapporté.

CONCLUSION Il faudra établir des critères diagnostiques pour l'ankyloglosse si on veut mener des études comparatives sur le traitement. La frénotomie est probablement un traitement efficace, mais il faut d'autres études contrôlées randomisées pour le confirmer. Il faut aussi élaborer des règles fiables pour prendre la décision de procéder à une frénotomie.

This article has been peer reviewed.
Cet article a fait l'objet d'une révision par des pairs.
Can Fam Physician 2007;53:1027-1033

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The effect of ankyloglossia on breastfeeding has been a matter of controversy in the medical literature for 50 years.[1] With the resurgence of breastfeeding, ankyloglossia has once again become an important clinical issue.[2-6] The prevalence of ankyloglossia has been reported in several studies,[5-9] but there is neither an accepted criterion standard nor clinically practical criteria for diagnosing the condition.[9] This lack of standardized criteria for diagnosing ankyloglossia is one of the Canadian Paediatric Society's main criticisms of research on this condition.[10]

Ankyloglossia in children can lead to a range of problems, such as difficulties breastfeeding, speech impediments, poor oral hygiene, and being embarrassed by peers during childhood and adolescence. About 90% of pediatricians and 70% of otolaryngologists believe that ankyloglossia rarely causes feeding difficulties; about 69% of lactation consultants believe that it frequently causes feeding difficulties, and an additional 30% believe it occasionally causes feeding difficulties.[11] Ankyloglossia in infants is associated with a 25% to 60% incidence of difficulties with breastfeeding, such as failure to thrive, maternal nipple damage, maternal breast pain, poor milk supply, breast engorgement, and refusing the breast.[2,3,7,8,12,13] Studies have shown that, for every day of maternal pain during the initial 3 weeks of breastfeeding, there is a 10% to 26% risk of cessation of breastfeeding.[14] The ineffective latch caused by ankyloglossia could be one of the primary underlying causes of all of these problems.[2,3,8,12,13]

Infants with restrictive ankyloglossia cannot extend their tongues over the lower gum line to form a proper seal and must use their jaws to keep the breast in the mouth.[2,3,8] An ultrasound study of breastfeeding in normal infants demonstrated that good tongue mobility is necessary for effective breastfeeding.[15] In infants with ankyloglossia, this deficiency cannot be ameliorated by the usual positioning and latching techniques and might require surgical correction.[2]

The most common treatment of infant ankyloglossia is simple frenotomy. Frenotomy is accomplished by incising several millimeters into the lingual frenulum. This procedure is brief and usually bloodless and is described in detail in a recent position paper from the American Academy of Pediatrics on the effect of tongue-tie on breastfeeding.[4] Hemostasis, if needed, is achieved by breastfeeding, which also lengthens the tongue and acts as an analgesic and antiseptic.[2]

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Complications historically attributed to frenotomy include infection, hemorrhage caused by severance of the lingual artery, and asphyxia caused by the released tongue falling back into the airway.[2,16] In recent years, there has been a renewal of interest in frenotomy as a treatment for ankyloglossia and an exploration of the complications associated with the procedure in the modern era, which are negligible.[5,6,9] Given the current revival of interest in frenotomy, we thought a comprehensive review of the literature on ankyloglossia and frenotomy would be helpful. As this study is a systematic review of the literature and did not involve direct acquisition of patient data or affect patient care, ethics approval was not required.

DATA SOURCES

MEDLINE and CINAHL databases were searched from 1966 to 2006 using combinations of the key words *ankyloglossia, tongue-tie, frenotomy, breastfeeding, breastfeeding problems, breastfeeding duration, latch, nipple trauma, pain, infant,* and *weight*.

Abstracts of articles related to ankyloglossia were retrieved and read by 1 of the authors (L.M.S.). The reference sections of selected papers were examined to identify further relevant articles. Hazelbaker's unpublished thesis[17] was acquired by the authors.

We selected articles we deemed were relevant to diagnosis, prevalence, and surgical treatment of ankyloglossia. These articles were read by 2 of the authors (L.M.S. and P.F.). These authors independently abstracted data, discussed discordances, and reached consensus on the results.

All studies that included data on infants younger than 6 months, such as case reports, case series, retrospective studies, prospective controlled studies, and randomized controlled trials, were selected for detailed analysis. Articles that did not address ankyloglossia in relation to breastfeeding problems were excluded. Opinion pieces, literature reviews, studies without patient data, studies that did not focus on breastfeeding, position statements, and surveys were also excluded. We used modified criteria[18] to evaluate the quality of the 6 studies addressing the effectiveness of frenotomy for treatment of ankyloglossia (**Table 1**).

SYNTHESIS

We found 183 articles during our initial search; 53 addressed ankyloglossia specifically. After exclusion criteria were applied, 5 studies describing the prevalence of ankyloglossia, 12 articles assessing diagnostic criteria for ankyloglossia, and 7 articles describing the effectiveness of frenotomy for treatment of ankyloglossia remained.

Table 1. Point system for assessing quality of the 6 studies that addressed the effectiveness of frenotomy for treatment of ankyloglossia

STUDY POPULATION
Patient selection 0—Not described 1—Any reason given 2—Only exclusion criteria given 3—Both inclusion and exclusion criteria given
Study design 1—Case report 2—Case series 3—Cohort study 4—Retrospective case-control study 5—Prospective case-control study 6—Randomized controlled trial
Sample size 1—<100 2—100-1000 3—>1000
Number of patients with ankyloglossia 1—<20 2—20-100 3—>100
Number of patients having frenotomy 0—0 1—<10 2—10-100 3—>100
Patient characteristics at baseline (1 point per criterion) • Age • Sex • Severity of tongue-tie • Severity of nipple pain • Severity of nipple trauma • Infant well-being (weight gain) • Latch or suck evaluation
Drop-outs 0—Not reported 1—Reported, drop-outs >0 2—Reported, drop-outs = 0 3—Reported with reason given
Losses to follow up 0—Not reported 1—Reported and >20% 2—Reported and <20%
INTERVENTION
Description of treatment (1 point per criterion) • Type of surgery: clipping or z-plasty • Anesthesia or not • Follow-up assessment of breastfeeding
MEASUREMENT OF EFFECT
Outcome measures (1 point per criterion) • Duration of breastfeeding • Complications • Latch or suck evaluation • Breastfeeding pain or trauma • Objective assessment of tongue mobility • Speed of infant weight gain
Duration of follow-up 1—<3 weeks 2—3-14 weeks 3—14 weeks-3 months
ANALYSIS AND RESULTS
Adequate analysis 0—None 1—Verbally described 2—Tabulated 3—Any statistical analysis 4—Formal systematic analysis
Adequate presentation of data 0—No data presented 1—Some data presented 2—All data presented

Diagnosis of ankyloglossia

Clinical criteria used to diagnose ankyloglossia are summarized in **Table 2**.[2,3,5-9,11,12,17,19-21] Criteria used for identifying ankyloglossia varied greatly from paper to paper. Many authors used criteria based on the physical characteristics of infants' oral anatomy. A commonly employed criterion was the frenulum being abnormally short and thick, which caused the tongue to become heart-shaped upon protrusion. Criteria also included signs of functional impairment, such as an inability to protrude the tongue past the gum line, and other indications of decreased tongue mobility. Some authors also cited the effect that ankyloglossia has on breastfeeding, such as causing maternal nipple pain and nipple trauma. None of these criteria have been validated. There is no accepted criterion standard test for ankyloglossia, and none of these studies prospectively compared its method against a proposed criterion standard. None of the studies assessed their diagnostic methods for internal and external validity.

Prevalence of ankyloglossia

The 5 studies that assessed the prevalence of ankyloglossia all used different diagnostic criteria and different ages of assessment for diagnosis (**Table 3**[5-9]). This could explain the variation in prevalence from 4.2% to 10.7%.

Consequences of ankyloglossia

Mothers breastfeeding infants with ankyloglossia have more nipple pain than mothers feeding normal infants.[7,9,18] The prevalence of nipple pain is between 60% and 80% in all nursing mothers during the early postpartum period.[13,22,23] With normal infants, this pain is transient, peaks on the third day, and resolves spontaneously within 2 weeks.[22] The prevalence of persistent nipple pain in breastfeeding women whose infants have ankyloglossia is between 36% and 80%. Only 3% of mothers of normal infants have intractable pain or difficulty getting their babies to latch at 6 weeks, but 25% of mothers of babies with ankyloglossia have these problems.[7]

Table 2. Criteria for diagnosing ankyloglossia

AUTHOR	YEAR	CRITERIA
Hogan et al[5]	2005	Frenulum extending along 25%–100% of tongues' total length
Ricke et al[9]	2005	Hazelbaker's assessment tool[17] for lingual frenulum function
Griffiths[6]	2004	Frenulum thick; tongue heart-shaped when protruded
Ballard et al[8]	2002	Hazelbaker's assessment tool[17] for lingual frenulum function
Messner et al[7]	2000	Frenulum abnormally short
Messner and Lalakea[11]	2000	Frenulum abnormally short; decreased mobility of tongue tip
Masaitis and Kaempf[3]	1996	Tongue heart-shaped when protruded; inability to bring tongue over lower gum ridge; abnormally short, thick frenulum; maternal nipple trauma
Harris et al[20]	1992	Frenulum short, thick, and fibrous; frenulum extends to the papillated surface of tongue
Marmet et al[2]	1990	Inability to bring tongue over lower gum ridge; normal breastfeeding sucking motion inhibited; tongue heart-shaped when protruded
Notestine[12]	1990	Frenulum <1 cm in length; tongue heart-shaped when protruded; tight feeling when finger placed under tongue along midline; tongue cannot reach gum line when protruded
Fleiss et al[19]	1990	Tongue tip cannot reach top of gums; tongue tip cannot swing from one corner of mouth to the other; tongue displays notching when protruded; tongue cannot be protruded beyond lower gum
Jorgenson et al[21]	1982	Frenulum prevents protrusion of tongue; frenulum extends to papillated surface of tongue; frenulum fissures tongue tip during normal movements

Table 3. Prevalence of ankyloglossia

STUDY	YEAR	N	COUNTRY	TYPE OF STUDY	DIAGNOSTIC METHOD	DAYS POSTPARTUM	PREVALENCE N (%) (95% CONFIDENCE INTERVAL)
Messner et al[7]	2000	1041	United States	Prospective trial	None; subjective impression with clinical correlate	Not applicable	50 (4.8) (3.6–6.3)
Ballard et al[8]	2002	3036	United States	Uncontrolled case series	Hazelbaker's assessment tool[17] for lingual frenulum function	2–3	127 (4.2) (3.5–5.0)
Griffiths[6]	2004	521	United Kingdom	Prospective uncontrolled cohort study	None; subjective impression with clinical correlate	18	All patients in study had ankyloglossia
Hogan et al[5]	2005	1866	United Kingdom	Randomized controlled trial	None; subjective impression with clinical correlate	3–70 (median 15)	201 (10.7) (6.6–14.74)
Ricke et al[9]	2005	3490	United States	Case-controlstudy	Hazelbaker's assessment tool[17] for lingual frenulum function	Both 1 and 30 days	148 (4.24) (1.1–7.42)

Frenotomy

Most of the 7 studies on frenotomy were of poor methodologic quality, with a mean quality score of 24.4 (range 9 to 40) out of a possible 47 points (**Table 4**[2,3,5,6,8,12,19]). The studies by Hogan et al,[5] Ballard et al,[8] Griffiths,[6] and Masaitis and Kaempf[3] had the highest scores. These 4 studies were larger and had more rigorous selection of patients and better descriptions of patients at baseline. There was only 1 randomized controlled trial of frenotomy.[5] All these studies used different outcome measures to assess the effectiveness of frenotomy, including nipple pain, infant growth, tongue mobility, and successful breastfeeding. All the studies showed an improvement in recorded outcomes after frenotomy.

In the randomized controlled trial,[5] 27 of 28 mothers of infants with ankyloglossia who were randomized to frenotomy had reduced nipple pain and improved breastfeeding at 1 week, but only 1 mother out of 29 randomized to 48 hours of intensive intervention by a lactation consultant experienced these improvements. All the remaining mothers in the control group (28/29) chose to have their infants undergo frenotomy. A second study[2] showed that intensive counseling and education on breastfeeding had not improved breastfeeding

difficulties at 2 to 12 weeks in 10 of 13 infants with ankyloglossia. In a prospective non-randomized cohort study,[6] 80% of infants had improved feeding 1 day after frenotomy.

None of the studies we found described serious complications following frenotomy. All the studies showed a benefit from frenotomy (**Table 5**[2,3,5,6,8,12,19]).

DISCUSSION

Our study has shown clearly that there is no accepted, widely used method for diagnosing ankyloglossia. The 1 standardized tool, the assessment tool for lingual frenulum function (ATLFF) developed by Hazelbaker,[17] is too

Table 4. Summary of assessment of research on ankyloglossia: *Best possible score was 47.*

CRITERIA	HOGAN ET AL[5]	BALLARD ET AL[8]	GRIFFITHS[6]	MASAITIS AND KAEMPF[3]	MARMET ET AL[2]	NOTESTINE[12]	FLEISS ET AL[19]
Study population							
• Drop-outs	3	2	2	1	2	NA	NA
• Losses to follow-up	2	2	0	0	NA	NA	NA
Intervention							
• Description of treatment	3	3	3	3	3	2	0
Measurement of outcome							
• Outcome measures	2	2	3	4	2	2	1
• Duration of follow-up	3	1	2	3	2	NA	NA
Analysis and results							
• Adequate analysis	4	3	3	2	2	0	1
• Adequate presentation of data	1	1	0	0	0	0	0
TOTAL	**40**	**31**	**31**	**28**	**21**	**11**	**9**

NA—Not applicable.

Table 5. Effectiveness of frenotomy for infants with ankyloglossia

STUDY	YEAR	N	TYPE OF STUDY	OUTCOME
Hogan et al[5]	2005	1866: 201 with ankyloglossia; 56 had frenotomy	Randomized controlled trial	54/56 had improved breastfeeding mechanics and reduced nipple pain with frenotomy; 79% improved immediately, and an additional 16% improved within 48 h
Griffiths[6]	2004	519 had frenotomy (215 were <3 mo)	Prospective uncontrolled cohort study	124/215 (57%) had improved feeding immediately; 174/215 (80%) had improved feeding at 24 h; 139/215 (65%) were still breastfed at 3 mo; 204/215 could extend tongue out of mouth at 3 mo
Ballard et al[8]	2002	3036: 123 with ankyloglossia; 35 had frenotomy	Uncontrolled case series	Decreased mean maternal pain score from 6.9 (± 2.31) to 1.2 (± 1.52); increased comfort for 31/35 breastfeeding mothers
Masaitis and Kaempf[3]	1996	2450: 36 had frenotomy	Case report	32/36 (89%) were breastfeeding 1 wk after procedure; 33/36 (92%) had normal tongue motion; 34/36 (94%) had appropriate growth at 3 mo; 36/36 (100%) reported normal criteria; 19/36 (53%) continued breastfeeding; 2/36 (0.5%) were weaned early due to breastfeeding problems
Marmet et al[2]	1990	13: 7 had frenotomy	Case report	5/7 had improved latch and decreased nipple pain and had resolved slow weight gain and milk-supply difficulties; 1/7 had improved suck dynamics; and 1/7 showed no improvement
Fleiss et al[19]	1990	3: 2 had frenotomy	Case report	1 showed improved sucking, and weight gain normalized; 1 developed a lisp
Notestine[12]	1990	2: 2 had frenotomy	Case report	1 mother had increased nipple comfort and less nipple trauma; 1 had increased comfort, and nipple distortion was resolved

lengthy and complex for use in a busy clinic, and in 1 study, could not be used to evaluate more than 60% of infants being tested.[9]

Development of a concise, practical, standardized, validated tool for diagnosing ankyloglossia and a decision rule for frenotomy are important for further research. They would allow researchers to conduct further randomized controlled trials and also permit integrated analysis of data from these trials.

Not surprisingly the data on prevalence and treatment of ankyloglossia are also complicated by heterogeneity, not only in diagnosis but also in assessment of outcomes. Despite this complication, the data suggest that ankyloglossia is common, causes problems, and can be relieved in most cases by frenotomy.

The prevalence of pain in mothers breastfeeding infants with ankyloglossia is much higher than that reported in mothers breastfeeding normal infants and clearly presents a considerable problem in terms of continuing breastfeeding. Intensive breastfeeding support is often inadequate for relieving breastfeeding difficulties in babies with ankyloglossia.

Results of studies assessing the effectiveness of frenotomy showed that breastfeeding mechanics improved and maternal pain decreased after the procedure. None of the studies found any serious complications of frenotomy. Given the relatively high prevalence of ankyloglossia, the large proportion of mothers of these infants with nipple pain, the strong association between pain and stopping breastfeeding, and the generally acknowledged health risks associated with not breastfeeding, frenotomy could be of great use as a safe and effective early intervention for breastfeeding problems attributed to ankyloglossia.

Future research efforts should be aimed at establishing clinically practical and valid diagnostic criteria for ankyloglossia in infants. After these criteria are established, results of further randomized controlled trials would assist in deciding whether frenotomy reduces breastfeeding difficulties, increases duration and exclusivity of breastfeeding, and leads to improved growth in tongue-tied infants. The design of an ideal randomized controlled trial, however, would be limited by ethical constraints. It would be unethical to leave a control group of mothers of infants with ankyloglossia in pain given that current evidence strongly suggests that frenotomy would relieve their pain and that continued pain would put their infants at increased risk of premature weaning.

Limitations

This review is limited to articles available in English and indexed on MEDLINE and CINAHL and papers referenced in them. Limiting the search technique might have missed studies of non-surgical approaches to tongue-tie and might have led to an overestimation of the prevalence of breastfeeding difficulties associated with ankyloglossia. We recognize that the methodology of this comparative review is subjective, but without standardized diagnostic criteria, a more systematic review of the literature was not possible.

EDITOR'S KEY POINTS

- Ankyloglossia is a common finding in infants. Prevalence ranges between 4.2% and 10.7%.
- Mothers breastfeeding infants with ankyloglossia have more nipple pain than mothers breastfeeding normal infants. The prevalence of persistent pain in these women is up to 80%; about 25% have intractable pain or difficulty latching at 6 weeks.
- Studies show a positive benefit of frenotomy with no serious complications.

POINTS DE REPÈRE DU RÉDACTEUR

- L'ankyloglosse est fréquente chez le nouveau-né, sa prévalence étant de 4,2% à 10,7%.
- Les femmes qui allaitent un bébé avec ankyloglosse ont plus de douleur mamelonnaire que celles qui nourrissent un enfant normal. La prévalence de douleur persistante chez ces femmes peut atteindre 80% et dans 25% des cas, on observe des douleurs rebelles ou un verrouillage difficile à 6 semaines.
- Les études montrent que la frénotomie a des effets favorables sans complication sérieuse.

Conclusion

Given the evidence currently available, we propose that frenotomy be viewed as a safe, effective, and practical approach to treatment of breastfeeding difficulties in infants with ankyloglossia in whom alternative explanations for poor feeding and failure to thrive have been properly assessed.

Acknowledgment

This study was funded through the research and development funds of the Herzl Family Practice Center. We thank **Alison Glaser**, *a research assistant in the Department of Family Medicine at McGill University in Montreal, Que;* **Francesca Frati**, *an Information Management Consultant for the Department of Family Medicine at Jewish General Hospital in Montreal; and* **A.K. Hazelbaker** *for allowing us to use her thesis,* The assessment tool for lingual frenulum function (ATLFF): use in a lactation consultant's private practice.

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