

Tongue-tie and frenotomy in infants with breastfeeding difficulties: achieving a balance

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ABSTRACT

Aims Currently there is debate on how best to manage young infants with tongue-tie who have breastfeeding problems. One of the challenges is the subjectivity of the outcome variables used to assess efficacy of tongue-tie division. This structured review documents how the argument has evolved. It proposes how best to assess, inform and manage mothers and their babies who present with tongue-tie related breastfeeding problems.

Methods Databases were searched for relevant papers including Pubmed, Medline, and the Cochrane Library. Professionals in the field were personally contacted regarding the provision of additional data. Inclusion criteria were: infants less than 3 months old with tongue-tie and/or feeding problems. The exclusion criteria were infants with oral anomalies and neuromuscular disorders.

Results There is wide variation in prevalence rates reported in different series, from 0.02 to 10.7%. The most comprehensive clinical assessment is the Hazelbaker Assessment Tool for lingual frenulum function. The most recently published systematic review of the effect of tongue-tie release on breastfeeding concludes that there were a limited number of studies with quality evidence. There have been 316 infants enrolled in frenotomy RCTs across five studies. No major complications from surgical division were reported. The complications of frenotomy may be minimised with a check list before embarking on the procedure.

Conclusions Good assessment and selection are important because 50% of breastfeeding babies with ankyloglossia will not encounter any problems. We recommend 2 to 3 weeks as reasonable timing for intervention. Frenotomy appears to improve breastfeeding in infants with tongue-tie, but the placebo effect is difficult to quantify. Complications are rare, but it is important that it is carried out by a trained professional.

INTRODUCTION

There is renewed interest in the issue of tongue-tie (ankyloglossia) in newborns and the need for frenotomy. The current debate centres on the matter of tongue-tie and perceived difficulties with breast feeding. It is considered that tongue-tie impedes the baby's ability to latch on during feeding and that the mother experiences pain. The problems manifest themselves quickly after the birth, with the latching problems appearing over the first 24 h and nipple pain on the second day.¹

The main drivers of the current discussion concerning tongue-tie are lactation nurses, breast feeding support groups and mothers who have experienced difficulties. Lawson² points out that the emphasis is no longer on breast feeding

promotion but rather on early correction of problems that interfere with breast feeding. To date, paediatricians have had a peripheral role and limited input into the debate. Neither the Royal College of Paediatrics and Child Health nor the Faculty of Paediatrics, Royal College of Physicians in Ireland has adopted an official stance, but the latter has established a working party to inform on the issue. The Canadian Paediatric Society in a position statement on ankyloglossia and breast feeding stated that 'based on available evidence, frenotomy cannot be recommended'.³ The Dutch have abandoned frenotomy nationally.⁴ The Japan Paediatric Society has stated that ankyloglossia does not cause feeding difficulties and that frenotomy is not necessary in infancy.⁵ The National Institute for Health and Care Excellence guideline supports the use of frenulotomy, stating that there are no major safety concerns about the division of ankyloglossia and limited evidence that the procedure can improve breast feeding.⁶ The UNICEF Baby Friendly organisation recommends the use of the procedure if ankyloglossia is causing problems with feeding.⁷ The American Academy of Pediatrics in its section on breast feeding concludes that, when tongue-tie is symptomatic, it should be treated as early as possible to minimise breast feeding problems.⁸

It has been previously reported in a study of 1500 healthcare professionals that paediatricians were the least likely group to recommend surgery.⁹ The findings were that 90% of paediatricians and 70% of otolaryngologists believed that tongue-tie never, or rarely, caused a problem. Some believe that good outcomes following frenotomy are partially due to a placebo effect and that breast feeding can get better over time as the mother becomes more proficient. On the other hand, Messner *et al*¹⁰ found that most lactation consultants believed that tongue-tie is a cause of breastfeeding difficulties and could be solved by frenotomy. The current debate about the surgical treatment of tongue-tie must be set against the ethos, supported by all paediatricians, which is to avoid unnecessary operations in babies and children if possible. This is based on a belief that many conditions improve spontaneously over time as a child grows and develops. Over the last few decades there has been a reduction in tonsillectomies for tonsillitis¹¹ and grommets for secretory otitis media.¹² The non-invasive management of talipes using the Ponsetti technique has reduced the previous discomfort associated with surgery.¹³ Some clinicians are concerned that a more liberal approach to frenotomy may result in 'therapeutic creep', with significantly more babies being subjected to the procedure.



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One of the reasons that frenotomy causes so much debate is that, in a case of tongue-tie and breastfeeding difficulties, a decision has to be made quickly, within 2–3 weeks, in order for it to be beneficial. The assessment pathway needs to be clear and the surgical service needs to be responsive and available. With the current uncertainties this is often not the case.

Paediatricians need to be well informed about the indications for frenotomy in breastfed babies with tongue-tie in order to better advise parents. Good assessment and selection are important because 50% of breastfed babies with ankyloglossia will not encounter any problems.¹⁴ This point is important when evaluating the risk/benefit ratio of the procedure.

The aim of this review is to evaluate the effectiveness of surgical intervention of tongue-tie in breastfed infants.

METHODS

A structured clinical question was devised using the PICO (Population, Intervention, Comparison, Outcome) method:

What evidence is there for or against the use of frenotomy in babies with tongue-tie?

Population: infants with tongue-tie

Intervention: surgery—frenotomy

Comparison: placebo/sham/lactation support

Outcome: efficacy of surgical intervention for baby and mother.

The databases searched for relevant papers were PubMed, Medline, The Cochrane Library and the Web of Science. Based on their likelihood to produce hits, *Archives of Disease in Childhood*, *Pediatrics*, *Journal of Pediatrics*, *Acta Paediatrica Scandinavica* and *Journal of Human Nutrition* were further examined. The reference lists of retrieved papers were searched for additional citations. Professionals in the field were personally contacted regarding the provision of additional data.

Searching was restricted to English language articles or those translated into English. The search words were tongue-tie, ankyloglossia, frenotomy and frenulectomy.

The types of studies included were randomised controlled trials, case-control studies, case series, cross-sectional studies, case reports, opinion pieces and review articles.

The inclusion criteria were infants less than 3 months old with tongue-tie and/or feeding problems. The exclusion criteria were infants with cleft lip, cleft palate, oral anomalies and neuromuscular disorders.

The titles and abstracts of all reports retrieved through the search strategy were screened. Full copies of all studies deemed eligible for inclusion were obtained. If it was unclear from the abstract whether an article was potentially eligible for inclusion, a full text copy of the study was retrieved. The final decision to include or exclude a publication was based on its relevance to the issue of tongue-tie and breastfeeding difficulties. The level of assessment was as follows: meta-analysis, randomised controlled trials (RCTs), case series, case reports, commentaries and opinion pieces.

RESULTS

Prevalence

There is wide variation in prevalence rates reported in different series, from 0.02% to 10.7%.^{15–18} Most professionals would accept that the likely rate is 2–5%. The variation in prevalence is due to the lack of a uniform definition. Some assessments concentrate on the visual appearance of the frenulum and tongue while others place greater emphasis on the tongue function. The latter is more important because it is what will determine whether the baby is able to breast feed effectively.

It is more common in male infants in a ratio 1.5–2.6:1, with high family correlations. Most cases are sporadic but mutations in the T-box transcription factor TBX22 may cause a hereditary ankyloglossia with an association with cleft lip and palate.¹⁹ In relation to environmental factors, there is an increased association with maternal cocaine abuse.²⁰

Clinical and functional assessment of tongue-tie

For the busy clinician the key point to appreciate is that, in a case of tongue-tie, the frenulum is attached close to the tip of the tongue.²¹ The tongue appearance should be examined when the tongue is lifted.²² In rare cases the frenulum attachment is to the proximal tongue base which is shortened and this can cause similar restrictions in tongue movements. When examined visually, the 'free tongue' length in newborns should be >16 mm; measurements of <11 mm indicate moderate ankyloglossia and <7 mm indicates severe ankyloglossia.²³ In practical terms, however, this measurement is difficult to perform in a wriggling newborn.

Another measurement-based assessment has been proposed by Ruffoli *et al.*²⁴ They state that, in a normal infant, the frenulum length is >2 cm and the intrinsic distance >2.3 cm.

One should be familiar with the tools used to assess function when one is assessing studies on tongue-tie and effects of frenotomy. The most comprehensive clinical assessment is the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF).²⁵ It consists of five appearance criteria and seven function criteria each scored on a 2/1/0 system. Seven movements are evaluated: lateralisation, lift, extension, cupping, peristalsis, spread of anterior tongue and snap back. Tongue-tie is diagnosed if the appearance score is ≤8 or the function score is ≤11. A study of 58 infants found the tool to be highly reliable.²⁶ Subsequently a study of over 140 babies found that 55.2% were not categorised using the HATLFF score.²⁷ Ricke *et al* reported that the inter-rater agreement using HATLFF was moderate and many infants did not conform to the categories designed.²⁸ It has been reported that the last four function items do not have good inter-rater reliability.²⁹ Hazelbaker has pointed out that HATLFF is a screening tool and is insufficient to be used as a predictor of breastfeeding outcomes.³⁰

In determining whether or not there are difficulties with breast feeding, a number of objective tools are used. The LATCH score,³¹ which is similar in design to the APGAR score, consists of five items marked on a 0–2 basis. The items are latch, audible swallowing, type of nipple, comfort and hold positioning. The LATCH score was found by Riordan *et al*^{31a} to be a useful identifier for mothers at risk of early weaning. It provides a useful assessment of breastfeeding success and is used as an outcome in many studies.

The short-form McGill Pain Questionnaire (SF-MPQ)³² is used in many studies to rate the mother's nipple pain. It takes 2–5 min to administer and has three sections. It is a short version of one of the most globally recognised pain assessments.

The Infant Breastfeeding Assessment Tool (IBFAT)³³ has been noted to correlate with breastfeeding competence and maternal satisfaction with breast feeding. While the LATCH score includes maternal nipple pain, the IBFAT tool does not. One small descriptive study found that the IBFAT and LATCH tools were not sufficiently reliable at that stage in their development to be valid for clinical use. They recommended revision and retesting before use in clinical practice to identify breastfeeding mother–infant pairs who need intervention.³⁴

Efficacy of frenotomy

Four systematic reviews and five RCTs have assessed the efficacy of frenotomy. Webb *et al*³⁵ in their methodological review confined their analysis to the 20 studies that met level 4 evidence or above. The objective improvements following frenotomy were LATCH scores (3 studies), SF MPQ Index (2 studies), IBFAT (1 study), feeding characteristics (3 studies). The subjective parameters were improvement in the maternal perception of breastfeeding (14 studies) and maternal pain scores (4 studies). Ito³⁶ has recently published a systematic review that included 4 RCTs and 12 observational studies. The two most important outcomes emphasised in the review were latching and nipple pain. The conclusion was a moderate quality of evidence for the effectiveness of frenotomy.

Segal *et al*³⁷ in a methodological review included seven articles describing the effectiveness of frenotomy. They stated that most of the studies were of poor methodological quality, the mean quality score being 24.4 out of a possible 47 points, but on balance frenotomy is probably an effective treatment. None of the studies prospectively compared its method of diagnosis against a proposed criterion standard. Furthermore, the studies used different outcome measures including nipple pain, successful breast feeding, tongue mobility and infant growth. Nipple pain was constantly highlighted.

Finigan *et al*,¹⁶ in a systematic literature review consisting of 5 RCTs and 8 case studies, concluded that frenotomy appears to offer long-term breastfeeding improvement for more than 50% of cases. The blinding in the RCTs raised a number of reservations. It can fail if there is a small blood stain on the swab in the intervention cases and also the blinding can only be maintained for a short period of time.

A total of 316 infants have been enrolled in 5 frenotomy randomised trials and the main findings are summarised in [table 1](#).

Hogan *et al*³⁸ performed a study on 57 babies (40 breastfed and 17 bottlefed), in which the index group had an outpatient frenotomy and the control group had 48 h intense lactation support followed by the offer of a frenotomy. The researchers were blinded. A 96% improvement was demonstrated in the frenotomy group (n=27) and a 3% improvement was shown in the control group (n=1). At 48 h the control group was offered frenotomy, which they all accepted. The authors themselves stated that one of the limitations was the lack of an objective measure of improvement.

Dollberg *et al*³⁹ recruited 25 infants with ankyloglossia whose mothers had sore nipples. The sequences in the two groups were (1) frenotomy, breast feeding, sham, breast feeding; and (2) sham, breast feeding, frenotomy, breast feeding. Both mothers and caregivers were blinded. An immediate significant decrease was noted in the pain score but not in the LATCH score after frenotomy compared with the sham procedure.

Buryk *et al*⁴⁰ performed a single-blind controlled trial of 58 infants randomised either to frenotomy (n=30) or sham procedure (n=28). The outcome variables were nipple pain and the IBFAT tool. The issue of breastfeeding duration could not be answered because all but one of the mother/baby dyads in the sham procedure limb had a frenotomy performed at 2 weeks.

A double-blind trial by Berry *et al*⁴¹ randomised 57 babies to division or non-division. They found that 78% (21/27) of the mothers in the division group reported an immediate improvement in breast feeding compared with 47% (14/30) in the non-intervention group. This is less striking than the results of non-blinded trials, perhaps reflecting the placebo effect. After the intervention, those infants allocated to non-division had their tongue-tie divided.

Emond *et al*¹⁷ in a study of 107 infants with mild to moderate tongue-tie compared frenotomy with controls. Those with severe tongue-tie were not included as they were offered immediate frenotomy. The primary outcome was the LATCH score at 5 days. The main finding was improved self-efficacy with feeding. The obvious reservation with the study was the omission of infants with severe tongue-tie. Longer follow-up was not possible because many of the mothers in the control limb opted for frenotomy after the end of the 5 day period.

There are a number of case-control studies. Ballard *et al*¹ devised a study in which the breastfeeding mothers acted as their own controls. Frenotomy was performed on 123 infants resulting in improved latch in all cases and maternal pain was significantly reduced. Ricke *et al*²⁹ enrolled 49 infants with tongue-tie and 98 control infants. All the infants were breastfed. Both groups were followed up and assessed at 1 week and 1 month. At 1 week the tongue-tie group was three times more likely to be bottle feeding (RR 3.11 CI 1.21 to 8.03), but they pointed out that 80% did successfully breast feed. At 1 month tongue-tie babies were as likely as controls to be bottlefed only. The authors did not find the ATLF useful in determining which tongue-tie infants would develop breastfeeding problems. Messner *et al*¹⁰ undertook a case-control study of 41 infants; 83% of tongue-tie infants and 92% of control infants were still breast feeding at 2 months.

Schwartz *et al* have reported that, for every day of maternal pain during the initial 3 weeks of breast feeding, there is a 10–26% risk of cessation of breast feeding.⁴² Geddes *et al*⁴³ recruited 24 mother-infant pairs experiencing breastfeeding problems. A combination of clinical and submental ultrasound parameters were measured before and after frenotomy. All clinical measures improved following the procedure. Before the surgery the ultrasound showed that the infants compressed either the tip or the base of the nipple. After the frenotomy both these ultrasound patterns resolved.

Sethi *et al*⁴⁴ reported an improvement in breast feeding at 2 weeks after frenotomy in 77% of mothers but not in the other 23%. The authors point out that tongue-tie is not the only breastfeeding problem encountered in these infants. Steehler *et al*⁴⁵ followed up 302 infants who underwent frenotomy for ankyloglossia. 80.4% of mothers strongly believed that the procedure benefited their ability to breast feed. Ochi⁴⁶ surveyed 20 mothers of infants with ankyloglossia before and 2 weeks after frenotomy; 15 controls that also had their score measured at a similar time interval. The breastfeeding symptom score decreased in the frenotomy group but remained unchanged in the control group. The reservation of the study is that neither the patients nor the doctors were blinded.

Hall and Renfrew¹⁴ in a comprehensive review almost 10 years ago made many of the points that are still valid today. Maternal pain during feeding and difficulty in latching the baby to the breast are the main problems attributed to tongue-tie. They point out that these difficulties can be caused by factors unconnected with tongue-tie. It is difficult get an objective assessment measure after frenotomy because, in many studies, the primary outcome measure is pain during breast feeding.

Isaacson *et al*⁴⁷ in their review of ankyloglossia point out that there is still a lack of consensus among clinicians about frenotomy. They suggest that, if there are continuing problems with breast feeding despite lactation support, frenotomy should be considered.

Table 1 Randomised trials of frenotomy

Citation	Study group	Study type	Primary outcome	Key result	Comments
Hogan <i>et al</i> ³⁸	57 babies randomised: 28 immediate division, 29 controls (lactation support)	RCT	Improvement in feeding	Division improved feeding (maternal gauge) in 96.42% (27/28) babies compared with lactation support alone ($p<0.001$)	All 28 of control group requested division at 48 h. Lack of objective measure of improvement
Dollberg <i>et al</i> ³⁹	25 mother/baby dyads Two sequences: 1. frenotomy, breast feeding, sham, breast feeding (n=14) 2. sham, breast feeding, frenotomy, breast feeding (n=11)	Randomised prospective cohort	Standardised LATCH score Standardised pain score	Non-significant improvement in LATCH score ($p=0.06$) Significant reduction in pain score ($p=0.001$)	All babies had frenotomy procedure as part of sequence
Buryk <i>et al</i> ⁴⁰	58 mother/infant dyads with maternal nipple pain or difficulty breastfeeding with significant ankyloglossia as judged by HATLFF: 30 frenotomy 28 sham	Single-blind RCT	SF-MPQ score IBFAT score	SF-MPQ scores reduced from 16.77 (SD1.88) before, to 4.9 (SD1.46) after intervention in frenotomy group. ($p<0.001$) IBFAT scores improved in the frenotomy group and were unchanged in sham group ($p=0.29$)	All but one parent in the sham group elected to have frenotomy at or before 2-week follow-up
Berry <i>et al</i> ⁴¹	60 breastfed babies: 27 division group 30 non-division group	Double-blind RCT	Maternal improvement in feeding Observer improvement in feeding	Significant immediate improvement in maternal gauge in 21 (78%) of division group compared with 14 (47%) of comparison group ($p<0.02$; 95% CI 6% to 51%) The results were reported in the study as follows: 78% (21 of 27) of mothers in Group A reported an immediate improvement in feeding following the intervention, compared with 47% (14 of 30) in Group B (two-tailed χ^2 $p<0.02$; 95% confidence interval, 6–51%).	Only objective observed change was better latch After the intervention, those infants allocated to non-division had their tongue-tie divided
Emond <i>et al</i> ¹⁷	107 infants with mild to moderate tongue-tie diagnosed by HATLFF-short form tool: 55 intervention group 52 comparison group	Randomised parallel group single-centre feasibility trial	LATCH score at 5 days	No difference in primary outcome (ie, LATCH score) between intervention and control groups at 5 days (0.52)	Severe tongue-tie was not included. Long-term follow-up not possible as only 8 (15%) of comparison group had not had a frenotomy at 8 weeks

HATLFF, Hazelbaker Assessment Tool for Lingual Frenulum Function; IBFAT, Infant Breastfeeding Assessment Tool; RCT, randomised controlled trial; SF-MPQ, short-form McGill Pain Questionnaire.

Timing of frenotomy and use of analgesia

It is evident from the studies that the breastfeeding difficulties in infants with ankyloglossia become apparent in the first few days after birth. Nipple pain and difficult latching lasting for >3 weeks after birth result in a 10–26% cessation in breast feeding.³⁴ The timing of the frenotomy across a series of studies was as follows: 6 days,⁴⁸ 1–21 days,³² 20 days,¹⁴ <2 weeks,³⁹ 18 days.¹⁴

In young infants frenotomy is usually performed at the outpatient clinic. When the infant is <3 months old it is undertaken without anaesthesia.³⁴ Some operators administer either sucrose or paracetamol. Griffiths⁴⁹ in a study of 200 infants undergoing frenotomy without analgesia found that 18% cried during the procedure and 60% after the procedure. The mean crying time was 15 s. A questionnaire study⁵⁰ has reported that frenotomy was safe with no or local analgesia. Ovental *et al*⁴⁸ in an RCT of topical benzocaine in 21 infants found that it was not beneficial. The average crying times in the benzocaine and control groups was 21.6 s and 13.1 s. Barrington⁵¹ states that some form of analgesia such as sucrose should always be administered to the infant.

Complications of frenotomy and their prevention

The effective way to minimise complications associated with the procedure is to ensure that the operator is skilled and properly trained. This is not always the case. A survey of 425 North American physicians⁶ reported that 22% had performed the

procedure but only 10% had received formal training. An Australian study⁵² reported 11 midwives trained in the technique of frenotomy over 5 years, and further staff being recruited and trained.

Complications following frenotomy are uncommon.^{53 54} The commonest is bleeding which usually stops quickly with local pressure. A local haematoma is another potential risk.⁵⁵ Care needs to be taken to avoid the lingual vein. Damage to the salivary ducts is a rare complication. Subacute massive oedema of the submandibular has been reported.⁵⁶ Two per cent of infants⁴⁹ developed an ulcer under the tongue and 2.6% of infants required a repeat procedure.¹⁵

The complications of frenotomy may be minimised with a check list before embarking on the procedure.

- ▶ Assessment by a lactation nurse to confirm that the tongue-tie is the cause of the breastfeeding difficulties.
- ▶ Oral and systemic examination by a paediatrician or GP to exclude other causes of poor feeding such as a urinary tract infection.
- ▶ Confirmation that the infant received vitamin K after birth.
- ▶ Note any bleeding disorders in the family.
- ▶ Parents should sign a consent form and be informed of the potential complications of the procedure.
- ▶ Ensure that the frenotomy is undertaken by an appropriately trained professional.
- ▶ Follow-up of the infant; if there is no improvement there may be another medical cause for the feeding problem.

CONCLUSIONS

We suggest that paediatricians, irrespective of their professional and personal views, should be in a position to inform and direct mothers appropriately when the issue of tongue-tie division arises in the early days after birth. Increasingly, patients are looking for additional information in order to be able to make an informed decision. They want to know what their options are.

Our review of the literature suggests that there is a wide variation in the prevalence of tongue-tie as it is not easy to measure objectively. The problem will usually present within the first week after birth, however its effect on feeding is difficult to gauge. If assessment indicates that tongue-tie is the causative factor, the dilemma is how long to wait. Sometimes, if one waits, the problem settles spontaneously. On the other hand, if one waits too long the pain and poor feeding may cause the mother to abandon breast feeding. We feel that a reasonable compromise is to intervene between 2 and 3 weeks of age. The evidence is complicated by the placebo effect, which is difficult to quantify. One of the most important unanswered questions in all the studies is whether the division of tongue-tie extends the duration of breast feeding, as in most of the studies the control group was offered frenotomy within a matter of days. The evidence predominantly suggests a subjective maternal improvement in breast feeding in infants with difficulty in breast feeding attributable to tongue-tie. Complications are rare, but it is important that the procedure is carried out by a trained professional who, in particular, is able to control any bleeding that may occur.

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