

# The effectiveness of frenulotomy on infant-feeding outcomes: a systematic literature review

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## Abstract

**Background.** Tongue-tie is a congenital anomaly in which the short lingual frenulum or highly attached genioglossus muscle restricts tongue movement. NICE (2005) called for robust clinical trials to be conducted on frenulotomy, the surgical procedure in which the lingual frenulum of infants is separated. The available evidence showed that the procedure posed very little, if any, risk for infants, yet NICE also noted that there was little evidence to underpin the conclusions being drawn by experts that frenulotomy improved infant-feeding.

**Aim.** To assess the effectiveness of frenulotomy on breastfeeding outcomes and maternal satisfaction with infant-feeding.

**Method.** The search was undertaken through Cochrane Library, Medline, CINAHL and UpToDate. Inclusion and exclusion criteria were set, and the search was restricted to evidence following the release of the 2005 NICE review. MESH search terms were applied: tongue-tie, ankyloglossia, frenotomy, frenulotomy, and breastfeeding. Two independent reviewers appraised all items, referring to the Cochrane Risk of Bias methodology. The Cochrane Library Critical Appraisal Skills Programme (CASP) tools and guidelines were used.

**Results.** A total of 53 papers were identified, of which 16 met the criteria: nine case studies, five randomised controlled trials, one prospective inter-rater reliability study and one systematic literature review. Frenulotomy appears to offer long-term breastfeeding benefits for more than 50% of cases. No evidence was available relating to the impact of tongue-tie on bottle-feeding infants.

**Implications.** Development of a robust, objective manual to assess the degree of tongue-tie and impact on feeding is vital. Research is needed to establish the impact on early parent-infant relationships of feeding a tongue-tied infant.

**Key words:** Tongue-tie, ankyloglossia, frenulotomy, breastfeeding, bottle-feeding, evidence-based midwifery

## Introduction

Partial ankyloglossia (tongue-tie) is defined as a short lingual frenulum that results in restricted movement of the tongue (Hazelbaker, 2010). The restriction appears to be in the forwards protrusion of the tongue (extension); lateral mobility (lateralisation) and tongue lift (Geddes et al, 2008). The reported incidence ranges from 3.2% to 10.7%, depending on the diagnostic criteria used at assessment (Messner et al, 2000; Ballard et al, 2002). There is a greater incidence in boys, with a male to female ratio of 1.5-2.6:1 (Renfrew, 2012). Anterior ankyloglossia is most common, being easily seen near the tip of the tongue and lending a classic heart-shaped appearance to the tongue. It often restricts extension, lift and lateralisation. The less common posterior ankyloglossia is often missed on examination, lying at the base of the tongue. Nevertheless, its functional effect also causes problems with feeding, particularly causing nipple pain and trauma (Hazelbaker, 2010). The findings of recent studies suggest that this condition is associated with feeding problems in early infancy and also with mastication, swallowing, dental and speech problems later in infancy (Geddes et al, 2008).

Historically, division of the lingual frenulum was performed routinely and early in infancy in order to prevent challenges with breastfeeding. The procedure was performed swiftly by the midwife (Notestine, 1990). There is convincing evidence that breastfeeding prevents infection, allergies and obesity in infants (Horta et al, 2007; UNICEF, 2012). It also impacts on maternal health and wellbeing, reducing the risks of breast and ovarian cancer (UK National Case Control Study Group, 1993; Furberg et al, 1999; Marmot et al, 2007), uterine inertia (Riordan, 2005) and osteoporosis (Chantray et al, 2004).

The purpose of this systematic review was to assess the effectiveness of frenulotomy on breastfeeding outcomes and maternal satisfaction with feeding infants.

## Rationale

In order to feed from the breast, a baby must be able to extend its tongue across its lower gum margin, cup the areola with its tongue, and elongate its mother's nipple to extend to the junction of its hard and soft palate (Geddes et al, 2008). The position of the tongue protects the delicate maternal nipple from trauma and enables the baby to move its tongue and jaw

freely to extract milk from the ducts within the breast (Geddes et al, 2008).

The symptoms reported by mothers of tongue-tied babies are clearly similar to the symptoms reported in the infant-feeding surveys (Bolling et al, 2007; Renfrew, 2012) that caused mothers to stop breastfeeding prematurely. These were detailed as:

- Sore, cracked and bleeding nipples
- Engorgement
- Recurrent blocked ducts
- Mastitis
- Breast abscesses
- Poor milk supply
- Unsettled baby
- Frequent feeding
- Faltering infantile growth.

With the resurgence of breastfeeding in the UK, ankyloglossia has again become an important issue in breastfeeding success or failure. However, across the UK, services to divide a tight frenulum in infants are sparse. The lack of objectivity in available studies to support assessment, treatment and evaluation of ankyloglossia results in inadequately robust foundations to underpin future service development.

Breastfeeding is pivotal to infant and maternal health and wellbeing; a key component in enabling mothers to build reciprocal and meaningful relationships with their infants (UNICEF, 2012). Advances in neuroscience have shown that the close and intimate contact during breastfeeding episodes enhances infant brain development and thus impacts on later life outcomes (Gerhardt, 2004; UNICEF, 2013).

## Method

A systematic review of the literature 'is a well planned review to answer a specific research question using a systematic and explicit methodology to identify, select and critically evaluate the results of the studies included' (Brettell and Grant, 2004; Aveyard, 2007). The Cochrane Critical Appraisal Skills Programme (CASP, 2004) tool was used to ensure consistency in review of the evidence, and the *Cochrane handbook for systematic reviews of interventions* (Higgins and Green, 2011) guided efforts in risk of bias assessment.

### Search strategy

The search strategy involved interrogation of electronic databases: Cochrane Library, Medline, CINAHL, and UpToDate. Limits were applied to restrict the search to publications from 2005 (the year of the NICE review) to 2013, relating only to humans, in English, and with full text available. MESH terms relevant to tongue-tie division in infants were applied: 'tongue-tie', 'ankyloglossia', 'frenotomy', 'frenulotomy', 'breastfeeding' and 'bottle-feeding'. A further search of Google Scholar and bibliographies sought papers that might have originally been overlooked. A total of 53 papers were identified.

### Study selection

Two reviewers independently reviewed the initial selection, applying inclusion and exclusion criteria. Items in which either reviewer was uncertain, or on which the reviewers differed were discussed and reviewed jointly until agreement was reached. The inclusion criteria for studies were:

- Included infants aged from birth up to six months of age
- Addressed challenges with breastfeeding or bottle-feeding
- Reported means of measuring the frenulum
- Employed randomised controlled trial (RCT), systematic literature review (after 2005), or case controlled study.

Exclusion criteria were:

- Sample focused on children older than six months
- No focus on feeding problems
- Reporting only on non-measured and non-notable frenulum
- Case series of less than five infants
- Surveys or opinion-based papers
- Duplicate studies.

A total of 16 reports met the criteria and formed the final selection: nine case studies, five RCTs, one prospective inter-rater reliability study, and one systematic literature review. No papers were found that examined the impact of ankyloglossia on bottle-feeding, most likely because the mechanics of feeding from the bottle differ and it is less common to find bottle-feeding problems in tongue-tied infants.

### Data extraction and synthesis

Data were extracted from the studies according to study design, nature and size of sample, intervention, outcomes, and significance and confidence intervals (see [Table 1](#)). The quality of the studies was assessed using the CASP tool. Risk of bias was evaluated with consideration of sample, data collection, clarity of inclusion and exclusion criteria, outcome measures and reliability. The review also appraised outcome level for randomisation, blinding and withdrawals. Quality assessment of RCTs was based on the classification of 'risk of bias' (Higgins and Green, 2011). The domains included low risk for bias, high risk for bias, and unclear categories.

## Results

All studies, except for Dollberg et al (2011), employed consecutive sampling. The RCTs employed double-blinding (Amir et al, 2006; Dollberg et al, 2006; Berry et al, 2012), or single-blinding of mothers (Buryk et al, 2011) or researchers (Hogan et

al, 2005). Blinding with this procedure relies on comparable periods of time for removal of the baby from the mother, insertion of a swab under the tongue for both intervention and control group babies, and feeding after the procedure without examination of the mouth by mother or observer. Even in a well-planned and competently conducted trial, such as that by Berry et al (2012), blinding can fail when a small blood stain appears on the swab. Clearly, blinding can be maintained for only a short period.

Details of blinding were not always clear, resulting in a greater risk of bias. Of the five RCTs, only one study presented effect size calculation (Buryk et al, 2011). Confidence intervals varied as would be expected in case series of different sizes, with smaller samples leading to wider confidence limits. However, the results indicated that the effect would still be likely to be demonstrated in larger studies.

#### *Assessment of degree of tongue-tie*

Assessment of the degree of tongue-tie and impact on tongue function was mainly subjective, with the construction of questionnaires as a common approach to assessment in case studies. Three studies employed the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) (Hazelbaker, 1993) for a more objective measurement (Ricke et al, 2005; Amir et al, 2006; Buryk et al, 2011). The HATLFF tool was found to have moderate inter-rater reliability, and was found to be too difficult and complex for use in a busy clinic by Ricke et al (2005). Although Amir et al (2005) had previously used the HATLFF for assessment, Amir et al (2006) drew similar conclusions to Ricke et al, finding that HATLFF had increased reliability in assessing three functions of tongue mobility (extension, lateralisation and lift), but the four scores that related to sucking functions were not reliable (cupping, tongue spread, peristalsis, snap-back). Geddes et al (2008) also found HATLFF usage to be problematic. The findings suggest that there is still a need to develop a robust, simple-to-use, clinically practical, valid assessment tool.

LATCH (Jensen et al, 1994) is an instrument that relies upon observation and description of five characteristics of breastfeeding: latch, audible swallowing, type of nipple, comfort and hold (or positioning). There is evidence that increase in LATCH score correlates with increased maternal satisfaction with breastfeeding and with decrease in breastfeeding problems (Riordan et al, 2001). IBFAT, the Infant Breastfeeding Assessment Tool (Matthews, 1988), focuses on four elements of breastfeeding behaviour: readiness to feed, rooting, fixing and sucking. It also includes a measure of the mother's perception of feeding and satisfaction with this. Again, the score has been shown to correlate well with maternal satisfaction with breastfeeding (Lewallen, 2006).

#### *Outcomes*

Eight studies provided subjective outcome measures of frenulotomy: (Hogan et al, 2005; Amir et al, 2006; Wallace et al, 2006; Khoo et al, 2009; Miranda and Milroy, 2010; Agiris et al, 2011; Berry et al, 2012; Steehler et al, 2012), and seven studies provided objective outcomes using validated tools (IBFAT or LATCH) (Srinivasan et al, 2006; Geddes et al, 2008; Khoo et al, 2009; Ridgers et al, 2009; Miranda and Milroy, 2010; Buryk et al, 2011; Dollberg et al, 2011). The LATCH score was used pre- and post-treatment in three studies (Geddes et al, 2008; Srinivasan et al, 2006; Dollberg et al, 2006) and Geddes et al (2008) and Srinivasan et al (2006) showed significant improvements in breastfeeding ( $p < 0.05$  and  $p < 0.0001$ , respectively). Furthermore, Geddes et al (2008) also used sub-mental ultrasonography to demonstrate reduced nipple compression following tongue-tie division which clearly is the cause of maternal pain and poor milk transfer.

Only one study compared pre- and post-division IBFAT scores (Buryk et al, 2011). The mean score improved from  $9.3 \pm 0.69$  to  $11.6 \pm 0.81$  following tongue-tie division, compared to subjects receiving a sham procedure where no improvement was noted ( $p < 0.029$ ). In the same study, while Short Form McGill Pain Questionnaire (SF-MPQ) pain scores improved significantly in both groups, scores reduced significantly more in the frenotomy group ( $p < 0.001$ ). Miranda and Milroy's (2010) prospective study of 51 neonates found that tongue-tie division improved breastfeeding and also infant weight gain. Breastfeeding increased by 19% ( $3 \pm 0.3$  sessions/24h) and bottle top-ups decreased by 81% ( $7 \pm 0.6$  sessions/24h)  $p < 0.001$ . At two weeks post-treatment, 90% of infants had gained weight by centiles, 6% had remained unchanged, and 4% had fallen 7.5 centiles. Overall, there was a gain of  $15 \pm 1.2$  centiles from  $41 \pm 2.5$  to  $56 \pm 2.4$  ( $p < 0.001$ ).

Eight studies reported subjective outcomes for breastfeeding, with mothers stating whether there were any differences in their feeding. For example, Hogan et al (2005) reported that 24 mothers (85%) had experienced improved breastfeeding following frenulotomy, with only one mother in the non-treatment group reporting similar improvement ( $p < 0.001$ ). Berry et al (2012) saw a significant improvement in breastfeeding ( $n = 21$ , 78%) in the treatment group, whereas only 14 mothers (47%) in the control group reported similar improvements.

#### **Discussion**

There remains a need to develop an objective, clinically simple, robust and reliable tool for lingual frenulum assessment. This is supported by previous reviews (Suter and Bornstein, 2009). Without this, studies of ankyloglossia seem set to continue being limited in the ability to provide clear indications of the need for frenulotomy, and practitioners will continue to struggle to present convincing business cases for further service development. The outcomes of frenulotomy performed for breastfeeding problems are largely projected from the subjective experiences of mothers. Very few studies have attempted to use standardised, objective measures during pre- and post-operative assessment phases.

The HATLFF tool was found to have only moderate reliability for function assessments of ankyloglossia, and clinicians experienced in assessing tongue function may feel that it equates only to their current subjective assessments. Assessment using the tool may be cumbersome in the clinical setting. However, if practitioners' assessments are not conducted robustly,

they will not provide sufficiently for evidence-based decisions. Either alternative instruments are needed, or existing tools require further adaptation to be practical and rigorous.

The most important finding from the studies reviewed in this paper is that frenulotomy for neonates and mothers who are experiencing breastfeeding challenges provides significant improvements and supports continued breastfeeding. It improved maternal experience. Tongue-tie division appears to provide some long-term benefits, with more than 50% of the mothers still breastfeeding three months after the procedure.

Although not the focus of this review, it was clear that there was little high-quality evidence regarding the identification of clinically significant symptoms or the effect of tongue-tie on articulation of speech. Yet it has been postulated that when a frenulum extends to the tip of the tongue, it prevents tongue lift and is associated with difficulty in speech (Webb et al, 2013).

Limitations to the evidence base include the possibility of selection bias as parental consent decisions may be affected by extraneous factors. In many cases, the mother had attended a breastfeeding clinic or been advised by a breastfeeding specialist prior to referral. There may be a preponderance in study samples of mothers who were already more motivated to breastfeed and who were optimistic that frenulotomy would be effective.

### Conclusion

There remains a paucity of evidence to underpin clinical decision-making on the assessment and outcomes of frenulotomy for infants and their families. If improvement in the initiation and continuation rates of breastfeeding in the UK are to be supported, well-evaluated, efficient and effective services need to be provided locally, where infants can have their tongue function assessed objectively and the frenulum divided, and where their mothers can access specialist support to continue to breastfeed.

There is also little evidence to support understanding of the impact of a tight frenulum on bottle-feeding infants. This warrants further investigation so that services are equitable for all mothers and babies. The experiences of parents feeding tongue-tied infants need to be accounted for as this may inevitably affect parents' building of early relationships with their offspring.

This review concludes that tongue-tie division is important in supporting both the initiation and continuation of breastfeeding, protecting the maternal nipple from trauma, ensuring effective transfer of breastmilk to the infant, and improve satisfaction in feeding for parents. Tongue-tie division has been reported to be a safe and simple procedure (NICE, 2005). More long-term studies are required to measure the short-, medium- and long-term effects of frenulotomy on breastfeeding and bottle-feeding, to investigate maternal satisfaction with feeding and to develop a robust and valid assessment tool.

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