

Does frenotomy improve breastfeeding problems in neonates with ankyloglossia?

Breastfeeding is the natural way of providing neonates with all the nutrients they need for growth and development; with exclusive breastfeeding recommended for the first 6 months of life (World Health Organization (WHO), 2017). In the UK, the breastfeeding landscape is complex: more mothers are breastfeeding but the continuation rates are amongst the lowest worldwide (UNICEF, 2017a). It is reported that 74% of women start breastfeeding their child from birth, but by 6–8 weeks only 47% of women are breastfeeding at all (partially or exclusively) (Public Health England, 2015). There is increasing national and global focus on improving breastfeeding rates due to the wide evidence of health, development and bonding benefits for mother and baby but also the social and economic benefits to the wider population (Department of Health and Social Care (DHSC), 2009; Department of Education and DHSC, 2015; UNICEF, 2017b). It has therefore been argued that, to avoid early cessation, efforts to promote breastfeeding should be refocused to include early correction of problems that interfere with breastfeeding (Lawson, 2014).

Ankyloglossia, commonly referred to as tongue-tie, is a congenital condition characterised by an abnormally short, thickened, or tight lingual frenulum that restricts mobility of the tongue (Garbin et al, 2013; Braccio et al, 2016). Tongue-tie can often be visually identified and this is often diagnosed as anterior ankyloglossia; however, appearance is not indicative of function (Dodds and Neiger, 2014). In cases where ankyloglossia cannot be visualised, it may be palpable as a tight midline filament in the soft tissue where the tongue joins the floor of the mouth, often described as posterior ankyloglossia (Garbin et al, 2013; Pransky et al, 2015). The procedure for the release of tongue-tie in neonates is known as frenotomy or frenulotomy, and involves the lingual frenulum being divided with sharp, blunt-ended scissors, usually performed without anaesthesia (Mettias et al, 2013; National Institute for Health and Care Excellence (NICE), 2005; Webb et al, 2013). Breastfeeding may be resumed immediately after frenotomy (NICE, 2005).

Abstract

Background There are global and national initiatives to improve breastfeeding rates and ankyloglossia appears to be contributing to breastfeeding cessation, despite significant controversy regarding the need for frenotomy and the success of the intervention on breastfeeding outcomes.

Aims To critically appraise contemporary literature regarding the effectiveness of frenotomy on reducing breastfeeding problems.

Methods An electronic literature search was systematically performed using CINAHL, PubMed and Cochrane databases. Back-chaining and a hand search of bibliographies were also used.

Findings Four key themes were identified from the literature; challenges diagnosing ankyloglossia including lack of a universally recognised assessment tool, apparent maternal breastfeeding improvements, breastfeeding improvements for the neonate, and issues with determining the longevity of breastfeeding.

Conclusion Overall the literature was of differing validity and applicability, therefore further longer-term RCTs are required with larger sample sizes to be able to account for cross-over of participants from comparison to frenotomy group. The evidence has shown that frenotomy may improve breastfeeding outcomes but since breastfeeding is a complex interaction between mother and baby, releasing ankyloglossia does not universally remove all breastfeeding problems.

Keywords

Breastfeeding | Infant feeding | Ankyloglossia | Tongue-tie

During normal suckling, a baby extends his tongue to the lower lip to create a seal along with his palate and cheeks, around the mother's breast; any oral cavity anomalies such as cleft palate may interfere with this

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seal (Jackson, 2012). Since ankyloglossia may affect the infant's ability to exhibit normal peristaltic motion, this physiological change can impact on breastfeeding ability, although it rarely causes any feeding problems to bottlefed infants, as bottle feeding requires little tongue muscle effort. Consequently, a mother experiencing breastfeeding difficulties with a tongue-tied baby is likely to switch to bottle-feeding as this may resolve her anxiety and feeding concerns (Geddes et al, 2008; Jackson, 2012).

Guidelines and standards for ankyloglossia

An accepted or clinically practical tool for diagnosing ankyloglossia does not exist (Mettias et al, 2013). Estimates of the prevalence of ankyloglossia therefore vary between 4–16%, depending on the population and diagnostic criteria used, with a 2–3:1 male predominance (Ricke et al, 2005; Brookes and Bowley, 2014). In addition, ankyloglossia is associated with a 25–80% incidence of difficulties with breastfeeding (Suter and Bornstein, 2009; Mettias et al, 2013; Garbin et al, 2013). The effect of ankyloglossia on breastfeeding has long been a matter of controversy in the medical profession, as many tongue-tied babies do breastfeed successfully (Ricke et al, 2005). However, ankyloglossia may cause a range of problems, including nipple and breast pain, frustration and dissatisfaction with feeding, low milk supply and breastfeeding cessation, poor latch, slipping off the breast during feeds, ineffective milk transfer, inadequate weight gain and failure to thrive (Jackson, 2012; Mettias et al, 2013; Garbin et al, 2013; Dodds and Neiger, 2014; Henry and Hayman, 2014). Furthermore, there appears to be no data on the natural history of untreated ankyloglossia (Francis et al, 2015), although it has been suggested that it may cause issues with speech development (Brookes and Bowley, 2014).

The national guidelines for division of ankyloglossia for breastfeeding were published almost 12 years ago (NICE, 2005). The guidelines acknowledge limited evidence on this subject but state that there are no major safety concerns to frenotomy, and that the procedure may improve breastfeeding (NICE, 2005). Since the national guidelines were published, there have been numerous new research studies, and controversy still exists due to the lack of a standard definition and criteria for diagnosing ankyloglossia and a dearth of evidence of its impact on breastfeeding. It is important that new research is not viewed in isolation but is instead understood in context with other studies on this topic (Cluett and Bluff, 2005). This systematic literature review aims to critically evaluate the latest published research and assess its implications for midwifery practice (Rees, 2005; Polit and Beck, 2010; International Confederation of Midwives (ICM), 2013).

Method

Information sources

An electronic literature search was systematically performed using CINAHL, PubMed and Cochrane databases in January 2017. Back-chaining and a hand search of bibliographies were also used where necessary.

Search strategy

Searches were limited to peer-reviewed, English-language articles published in the past 10 years. Boolean operators were used, combining key word searches for variants of 'breastfeeding' and 'ankyloglossia'. The search results were then screened with a requirement to meet Level 4 evidence or above (Centre for Evidence Based Medicine (CEBM), 2009). *Figure 1* details the search and exclusion process.

Study selection

The author independently reviewed the list of titles and abstracts generated by the literature search for those meeting the aforementioned criteria.

The author, a midwife, chose to specifically focus on studies relating to the neonatal period, therefore excluding studies that included babies over 6 weeks of age at the time of frenotomy unless the mean or median age of the total sample was less than 6 weeks. Studies of ankyloglossia unrelated to breastfeeding problems were also excluded.

Critical review

The eight articles selected for review are summarised in *Table 1*. They were all appraised using tools from the critical appraisal skills programme (CASP) (2017a; 2017b; 2017c).

Design and methodology

Experimental designs, and randomised control trials (RCTs) specifically, are universally deemed the most scientifically valid method of research (Rees, 2005; CEBM, 2009); however, it is important to acknowledge that threats to validity exist (Campbell and Sandy, 1963). One such confounding variable not discussed in any of the research is whether the mothers had previous experience of breastfeeding and how, if at all, this influenced their decision, intent and experience of breastfeeding. It is worth noting that, in one study, there were 95 women who met inclusion criteria but declined to be randomised, and almost half wanted frenotomy immediately (Emond et al, 2014). Interestingly, these women had had a previous infant with ankyloglossia.

Maturation is also a threat to validity (Campbell and Sandy, 1963) in that the passage of time influences the change in pre- and post-intervention results due to normal physical, psychological and social changes

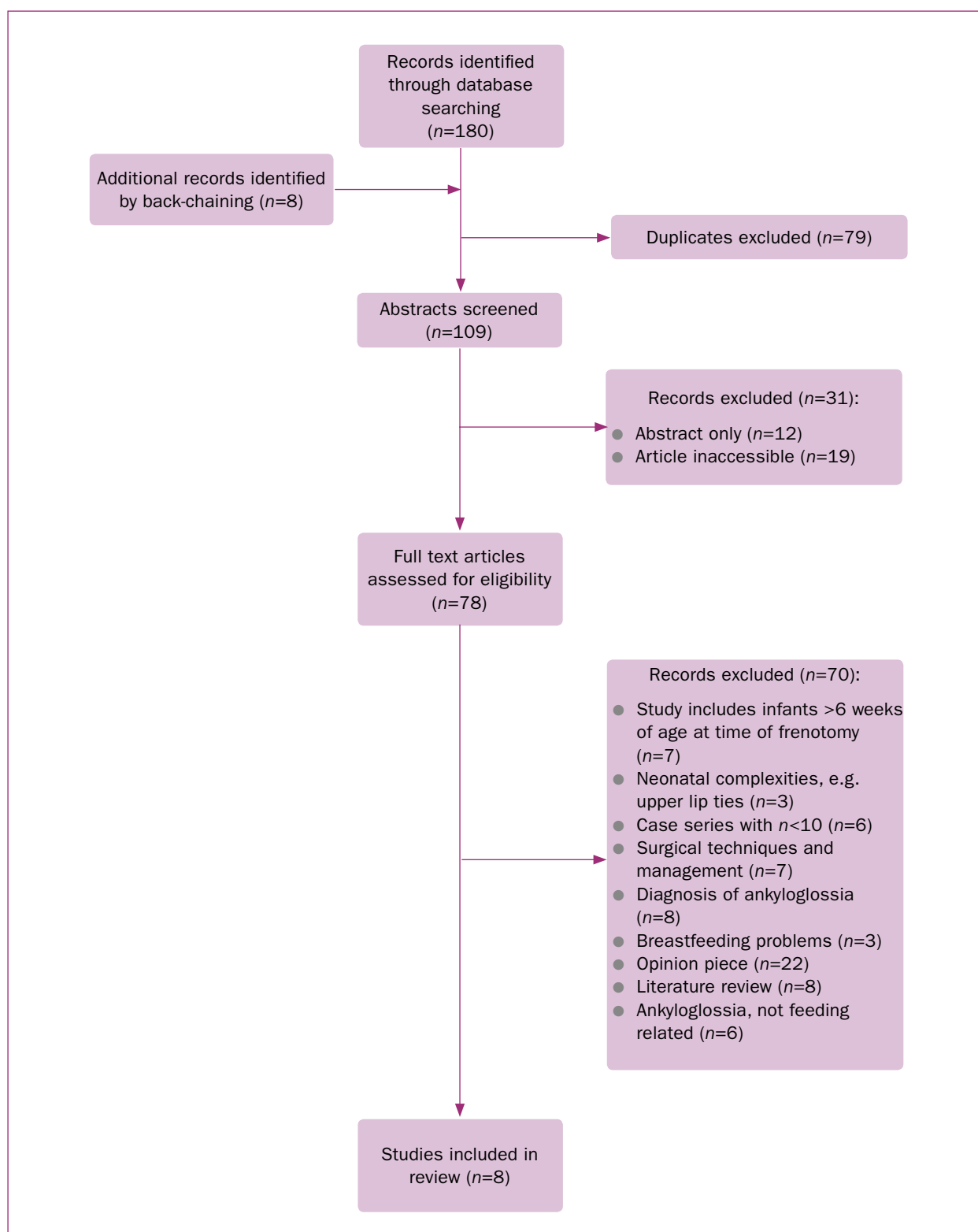


Figure 1. Flow diagram of the search and selection processes

of individuals, which are unrelated to variables in the study. Emond (2014) had an initial post-intervention assessment at 5 days and then a final 8-week follow-up, whereas Buryk et al (2011) had an initial assessment immediately after intervention, then 2-weeks later, and

additional regular follow-ups over a 1 year period. Hong et al (2010) used a retrospective record review without direct follow-up with the patients. The remaining studies had follow-ups ranging from 2 weeks to 3 months post-frenotomy (Khoo et al, 2009; Miranda and Milroy, 2010;

Table 1. Summary of studies included in literature review

Author	Country	Study type	Aim or Hypothesis	Sample	Outcome measures	Results
Buryk et al (2011)	USA	Randomised control trial (RCT)	To determine whether frenotomy improved nipple pain, ability to feed and length of breastfeeding	58 mother/infant dyads with significant ankyloglossia as judged by HATLFF. 30 frenotomy, 28 sham Age range = 1–35 days, Mean age = 6 days	SF-MPQ score IBFAT score	Frenotomy group more improved pain score than sham. Breastfeeding scores increased in frenotomy group. Unable to compare long term due to crossover
Emond et al (2014)	UK	RCT	To determine if frenotomy was better than standard breastfeeding support	107 mother/infant dyads with mild-moderate ankyloglossia using HATLFF. 55 intervention, 52 comparison group. Age range = 8–16 days Median age: 11 days	LATCH score IBFAT score BSES Pain VAS score	No difference in LATCH. Improved breastfeeding self-efficacy. Increased bottle feeding in comparison group at 5 days
Edmunds et al (2013)	Australia	Qualitative study	To understand breastfeeding experiences of women whose infants have tongue-tie	10 mothers (8 primiparous, 2 multiparous) Infant age range = 3–21 days	Focused interviews	A common tension between women's expectations and the breastfeeding challenges they faced
Hong et al (2010)	USA	Case series	To determine prevalence of anterior versus posterior ankyloglossia and assess outcomes post frenotomy	341 infants (227 male, 114 female) Age range = 1–168 days Median age = 2.7 weeks	Retrospective chart review	94% anterior, 6% posterior. Frenotomy improved breastfeeding
Khoo et al (2009)	UK	Prospective case series	To establish whether those experiencing nipple pain were more likely to benefit from frenotomy	62 mother-infant dyads Age range = <90 days, Mean age = 23.5 days	Devised questionnaire	Mothers experiencing nipple pain when breastfeeding were most likely to benefit from frenotomy
Miranda and Milroy (2010)	UK	Prospective case series	Assess impact of frenotomy on neonatal growth and breastfeeding	51 neonates Age range = 12–36 days	Devised questionnaire	Ankyloglossia may detrimentally affect neonatal growth and frenotomy provides significant improvement
Sethi et al (2013)	UK	Case series	Evaluate indications and outcomes of frenotomy performed in infants with ankyloglossia for breastfeeding difficulties	85 infants (52 successfully followed-up) Age range = 4–37 days, Mean age = 14 days	Telephone questionnaire	Lack of universal improvement suggests that ankyloglossia was not the only cause of breastfeeding problems
Steehler et al (2012)	USA	Cohort study and retrospective survey	Measure maternal breastfeeding benefit after frenotomy and if timing affects outcome	367 infants (n= 302, 82.3% frenotomy; n=65, 17.7% no intervention). Mean age = 18 days	Telephone interview	When frenotomy performed in first week of life there was more benefit than when it is performed after the first week of life

HATLFF: Hazelbaker Assessment Tool for Lingual Frenulum Function; SF-MPQ: Short form McGill Pain Questionnaire; IBFAT: Infant Breast Feeding Assessment Tool; LATCH: Latch, Audible swallowing, nipple Type, Comfort and Hold; BSES: Breastfeeding Self Efficacy Score; VAS: Visual Analogue Score

Edmunds et al, 2013; Sethi et al, 2013) with the exception of Steehler et al (2012), where, due to the nature of the retrospective survey, some participants were followed-up 5 years post-intervention. While Steehler et al (2012) found no clear trends on response directly linked to the duration between intervention and follow-up, it is important to acknowledge potential recall bias whereby participants reconstruct memories retrospectively, or

to fit the circumstances in which they are being asked (Gomm et al, 2000).

Sampling

A common sampling bias across all studies in this review was that all mothers were highly motivated to breastfeed. This is evidenced by their persistence to breastfeed, to seek help and support from midwives, GPs and lactation



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This study synthesised the literature to assess the effect of frenotomy on breastfeeding problems

consultants, and to consent to a surgical procedure for their infant to continue breastfeeding. In addition, Emond et al (2014) acknowledged that the mothers recruited in the trial had a higher education level and were less likely to come from a minority background compared with the general profile of women delivering at the trial hospital.

Five studies used cohort sampling, whereby a specific cohort was identified (namely, infants with ankyloglossia and breastfeeding problems within a specific time-frame) (Khoo et al, 2009; Hong et al, 2010; Miranda and Milroy, 2010; Steehler et al 2012; Sethi et al, 2013). The strength of using a cohort is that it reduces sampling error (Rees, 2005); however, since the inclusion criteria is a subjective measure reliant on health professional diagnosis, sampling error is likely to exist in these studies.

Edmunds et al (2012) used purposive sampling, which is a suitable choice for qualitative research as it ensures appropriateness and adequacy of the sample against the aims of the research (Morse and Field, 1996). Emond et al (2014) also used purposive sampling for the qualitative interviews following the initial RCT. This was to ensure a range of feeding methods and age of babies in both arms of the trial.

Participants in the study by Emond et al (2014) were randomised to control or intervention group and the researchers, but not the mothers, were blinded to the group assignment. In contrast, Buryk et al (2011) blinded the parents to the group to which their infants were allocated, but not the researchers. Parents were blinded only until initial post-procedure feeding as it was not

feasible to prevent mothers from looking in their infant's mouths. Both concealment of group allocation and blinding are important in assessing rigour of experimental designs (Rees, 2005), although Schultz (2001) suggests that concealment of group allocation from the subjects is more important than blinding in midwifery research, especially as the nature of some midwifery trials means that double-blinding is not possible. Blinding of the mothers is nevertheless essential to ensure the responses are not influenced by knowledge of group allocation.

Subject attrition

One study (Steehler et al, 2012) was impacted by loss of subjects to follow-up, with 367 infants included in the cohort study but only 91 (24.9%) mothers agreeing to participate in the telephone survey; a non-deliberate sampling bias. This limited the sample size of the non-frenotomy group to 9 participants. In another study (Buryk et al, 2011), significant crossover of sham to intervention group was experienced due to the nature of the subject: by the 2-week follow-up, all but one parent had opted for frenotomy and the study was therefore unable to address whether frenotomy increased longevity of breastfeeding. In addition, one-quarter of the subjects were lost to follow-up by the final 12-month review (Buryk et al, 2011).

Results and statistical analysis

All studies found improvements in breastfeeding outcomes to varying extents; some statistically significant and others not measured for statistical significance. Buryk

6 There is a wide variation in the prevalence of ankyloglossia, primarily due to a lack of a universally accepted definition and therefore no validated tool for diagnosis. 9

et al (2011) found the frenotomy group had significantly reduced pain than the sham group ($P < 0.001$) and that breastfeeding scores significantly improved in the frenotomy group ($P = 0.029$). Two further studies reported improved breastfeeding following frenotomy, but had no statistical significance in support of this claim (Hong et al, 2010; Sethi et al, 2013). One study (Emond et al, 2014) found that other breastfeeding measures and pain were no different from those in the comparison at 5 days, although the change in scores from 0–5 days was significantly greater in the intervention group ($P < 0.002$ and $P < 0.09$ respectively).

Thematic analysis

A number of themes arose throughout the review of the research literature, and were summarised in a matrix with relevant citations from each of the studies. A total of eight themes were present in two or more of the studies. It was decided that key themes would be selected if they appeared in three or more of the studies; resulting in four key themes to be explored in depth:

- Challenges diagnosing ankyloglossia
- Women's experiences of breastfeeding after frenotomy
- Neonatal experience after frenotomy
- Longevity of breastfeeding.

Challenges diagnosing ankyloglossia

A consistent theme among the studies was the issue of diagnosing and classifying ankyloglossia in infants with breastfeeding problems. Some women had tongue-tie identified by nursing staff, but it was rarely identified as a potential cause of breastfeeding problems (Edmunds et al, 2013). Two studies (Buryk et al, 2011; Emond et al, 2014) used the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF), but there were discrepancies in the interpretation and classification of the scores. It has been argued that the HATLFF is based on subjective assessments and requires expertise to apply it consistently (Amir et al, 2006), and that many infants do not conform to categories within the tool (Madlon-Kay et al, 2008). One study suggested that more than half (55.2%) of infants with ankyloglossia cannot be correctly categorised using the HATLFF score (Amir, 2006).

The remaining studies in this review used only subjective diagnosis by health professionals such as lactation consultants and paediatric otolaryngologists;

all using different techniques and classifications. There is no consensus regarding the precise definition and classification of ankyloglossia and several classification systems are available, but none correlate to severity of symptoms experienced when breastfeeding and so cannot be used as predictors of frenotomy success (Notestine, 1990; Kotlow, 1999; Hazelbaker, 2005; Amir et al, 2006). It is therefore important that more research is conducted to gain consensus for a precise definition of ankyloglossia to enable a suitable tool for universal diagnostics. Perhaps the creation of an international panel of practitioners would enable the construction of a definition and set of guidelines for researchers to produce comparable studies.

Women's experiences of breastfeeding after frenotomy

All of the research articles focused, with varying degrees of emphasis, on mothers' experiences of breastfeeding. Women described a release from pain when breastfeeding following frenotomy, which related not only to the physical pain but a release from the anxiety of wanting to breastfeed their baby but battling against pain symptoms (Edmunds et al, 2013). This supports the improvement in breastfeeding assessment following frenotomy (Khoo et al, 2009; Buryk et al, 2011), although Emond et al (2014) found no improvements in Latch, Hold, nipple Type, Comfort and Hold (LATCH) score. The majority of mothers believed the procedure benefited their child's ability to breastfeed (Miranda and Milroy, 2010; Steehler et al, 2012; Sethi et al, 2013). Furthermore, maternally rated self-efficacy has been shown to correlate well with overall duration of breastfeeding (Bandura, 1977; Baghurst et al, 2007).

Despite many women finding relief from painful feeding and improvements in their baby's feeding behaviours post-frenotomy, this did not apply to all, which highlights that there are other factors that need to be considered in the mother-infant breastfeeding dyad, and that frenotomy is not a one-size-fits-all solution. In particular, consistent counselling and support from skilled lactation consultants was shown to be imperative to better breastfeeding outcomes, as evidenced by lactation specialist involvement in all research studies in this review (Khoo et al, 2009; Hong et al, 2010; Miranda and Milroy, 2010; Buryk et al, 2011; Steehler et al 2012; Sethi et al, 2013). Furthermore, the lack of universal improvement suggests that ankyloglossia it is not the only cause of breastfeeding problems, and that frenotomy is not the only solution (Sethi et al, 2013).

Neonatal experience after frenotomy

In one study, all infants had anaesthetic before the procedure, and of those participants, 8 (2.6%) revision procedures were successfully performed for incomplete clipping or scarring (Steehler et al, 2012). Another study

noted 4 (4%) revision frenotomies, and 63 (64%) cases of a small white patch at the base of the frenulum, reported by the mother at 5 days, which took a median of 7 days to heal. No safety concerns were raised or reported in the remaining studies, and it is acknowledged that potential complications and damage to the submandibular duct are extremely rare (Masaitis and Kaempf, 1996).

A statistically significant reduction in length of feed and increase time between feeds ($P < 0.001$) were reported in one study (Khoo et al, 2009). Miranda and Milroy (2010) reported that the number of breastfeeding sessions in a 24-hour period significantly decreased by 19% at 2-weeks post procedure, and that there was an 81% improvement in the number of supplementary artificial feeds ($P < 0.001$), indicating more efficient feeds for the infant. This is consistent with other studies that have shown improved breastfeeding efficiency following frenotomy (Geddes et al, 2008).

Longevity of breastfeeding

Due to crossover in both RCTs of women in comparison groups requesting frenotomy (Buryk et al, 2011; Emond et al, 2014), accurate long-term measurement was not possible. It is not possible to maintain the control-group without crossover in a study of this kind without compromising the interests of participants (Rees, 2005; DHSC, 2005; 2011; ICM, 2014). Khoo et al (2009) reported a high rate of long term success, with 78% of mothers breastfeeding at 3 months, and Steehler et al (2012) found that 82.9% continued to breastfeed in the intervention group versus 66.7% in the no intervention group, although the time of measurement was not clear.

One study found that mothers presenting with nipple pain showed significant long term benefits from frenotomy and were more likely to be breastfeeding at 3 months following frenotomy (OR 5.8; 95% CI 1.1–31.6) (Khoo et al, 2009). Conversely, presence of a family history of ankyloglossia decreased the likelihood that a baby was breastfeeding at 3 months (OR 0.24; 95% CI 0.06–0.94) (Khoo et al, 2009). There is no evidence to suggest why family history has this effect, but sociocultural or other external influences may be a contributory factor in the complex mother-baby breastfeeding dyad. In short, more long-term research on longevity of breastfeeding after frenotomy is required.

Summary

This review of the literature suggests there is a wide variation in the prevalence of ankyloglossia, primarily due to a lack of a universally accepted definition and therefore no validated tool for diagnosis. The challenge is made more complex as the severity of tongue-tie does not correlate to severity of breastfeeding problems or likelihood of success of frenotomy. In addition, some

Key points

- There is a wide variation in the prevalence of ankyloglossia, primarily due to a lack of accepted definition and validated tool for diagnosis
- The issue is complex since the severity of ankyloglossia does not correlate to severity of breastfeeding problems or likelihood of success of frenotomy
- Many—but not all—women and infants experience improvements in breastfeeding problems and neonatal outcomes after frenotomy
- Any breastfeeding problems may cause women significant stress and anxiety; therefore the timing of any support or intervention is critical to reduce breastfeeding cessation rates
- This review has shown that frenotomy may improve breastfeeding outcomes but that breastfeeding is a complex interaction between mother and baby, with ankyloglossia being just one factor that may contribute towards problems

CPD reflective questions

- Estimates of the prevalence of ankyloglossia vary. What is the prevalence of tongue-tie in your setting?
- What evidence and/or guidelines are used in your setting to diagnose ankyloglossia and to advise women and families?
- Do you have experience of caring for babies with tongue-tie? Have you recorded an improvement in breastfeeding in case where frenotomy has been undertaken?

infants with ankyloglossia breastfeed successfully and there appears to be no data on the natural history of untreated ankyloglossia, although it has been suggested that it may cause issues with speech development. Many women and infants experience improvements in breastfeeding problems and neonatal outcomes following frenotomy, which is overall considered a safe procedure. It is therefore essential to acknowledge that frenotomy should be considered and offered, even if the improvement in outcomes is not universal.

A woman's desire to breastfeed should be encouraged due to the undisputed benefits of breastmilk, yet this is not always without difficulty. Support from lactation consultants is essential for initiating and maintaining good breastfeeding behaviours for women who are facing difficulties. Any breastfeeding problem may cause women significant stress and anxiety at a time of heightened emotion; therefore the timing of any support or intervention is critical to reduce breastfeeding cessation rates (although this was not explored in this article). Longevity of breastfeeding is ethically complex to measure as an RCT, as evidenced by the crossover from comparison to frenotomy groups in both RCTs in this review.

A possible limitation of this study is the number of articles that were inaccessible through the British Library Loan Service, particularly one RCT that appeared relevant (Berry et al, 2012). It would still be possible to

conclude that there is a good deal of available research on this subject, but it appears to be of limited-to-moderate value. More rigorous, longer term RCTs with larger sample sizes are required to be able to account for possible crossover and attrition, and give truly meaningful results. The evidence in this review has shown that frenotomy may improve breastfeeding outcomes, but it is also clear that breastfeeding is a complex interaction between mother and baby, and ankyloglossia is just one factor that may contribute towards problems. **BJM**

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