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Frenotomy for tongue-tie in newborn infants (Review)

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[Intervention Review]

Frenotomy for tongue-tie in newborn infants

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ABSTRACT

Background

Tongue-tie, or ankyloglossia, is a condition whereby the lingual frenulum attaches near the tip of the tongue and may be short, tight and thick. Tongue-tie is present in 4% to 11% of newborns. Tongue-tie has been cited as a cause of poor breastfeeding and maternal nipple pain. Frenotomy, which is commonly performed, may correct the restriction to tongue movement and allow more effective breastfeeding with less maternal nipple pain.

Objectives

To determine whether frenotomy is safe and effective in improving ability to feed orally among infants younger than three months of age with tongue-tie (and problems feeding).

Also, to perform subgroup analysis to determine the following.

• Severity of tongue-tie before frenotomy as measured by a validated tool (e.g. Hazelbaker Assessment Tool for Lingual Frenulum Function (ATLFF) scores < 11; scores ≥ 11) (Hazelbaker 1993).

• Gestational age at birth (< 37 weeks' gestation; 37 weeks' gestation and above).

• Method of feeding (breast or bottle).

• Age at frenotomy (≤ 10 days of age; > 10 days to three months of age).

• Severity of feeding difficulty (infants with feeding difficulty affecting weight gain (as assessed by infant's not regaining birth weight by day 14 or falling off centiles); infants with symptomatic feeding difficulty but thriving (greater than birth weight by day 14 and tracking centiles).

Search methods

We searched the Cochrane Central Register of Controlled Trials (CENTRAL), MEDLINE, Embase and CINAHL up to January 2017, as well as previous reviews including cross-references, expert informants and journal handsearching. We searched clinical trials databases for ongoing and recently completed trials. We applied no language restrictions.

Selection criteria

Randomised, quasi-randomised controlled trials or cluster-randomised trials that compared frenotomy versus no frenotomy or frenotomy versus sham procedure in newborn infants.

Data collection and analysis

Review authors extracted from the reports of clinical trials data regarding clinical outcomes including infant feeding, maternal nipple pain, duration of breastfeeding, cessation of breastfeeding, infant pain, excessive bleeding, infection at the site of frenotomy, ulceration at the site of frenotomy, damage to the tongue and/or submandibular ducts and recurrence of tongue-tie. We used the GRADE approach to assess the quality of evidence.

Main results

Five randomised trials met our inclusion criteria (n = 302). Three studies objectively measured infant breastfeeding using standardised assessment tools. Pooled analysis of two studies (n = 155) showed no change on a 10-point feeding scale following frenotomy (mean difference (MD) -0.1, 95% confidence interval (CI) -0.6 to 0.5 units on a 10-point feeding scale). A third study (n = 58) showed objective improvement on a 12-point feeding scale (MD 3.5, 95% CI 3.1 to 4.0 units of a 12-point feeding scale). Four studies objectively assessed maternal pain. Pooled analysis of three studies (n = 212) based on a 10-point pain scale showed a reduction in maternal pain scores following frenotomy (MD -0.7, 95% CI -1.4 to -0.1 units on a 10-point pain scale). A fourth study (n = 58) also showed a reduction in pain scores on a 50-point pain scale (MD -8.6, 95% CI -9.4 to -7.8 units on a 50-point pain scale). All studies reported no adverse effects following frenotomy. These studies had serious methodological shortcomings. They included small sample sizes, and only two studies blinded both mothers and assessors; one did not attempt blinding for mothers nor for assessors. All studies offered frenotomy to controls, and most controls underwent the procedure, suggesting lack of equipoise. No study was able to report whether frenotomy led to long-term successful breastfeeding.

Authors' conclusions

Frenotomy reduced breastfeeding mothers' nipple pain in the short term. Investigators did not find a consistent positive effect on infant breastfeeding. Researchers reported no serious complications, but the total number of infants studied was small. The small number of trials along with methodological shortcomings limits the certainty of these findings. Further randomised controlled trials of high methodological quality are necessary to determine the effects of frenotomy.

PLAIN LANGUAGE SUMMARY

Surgical release of tongue-tie for the treatment of tongue-tie in young babies

Review question: Tongue-tie is a potentially treatable cause of breastfeeding problems - if a baby is tongue-tied and is having feeding difficulties, does releasing the tongue-tie help?

Background: Tongue-tie is a condition whereby the membrane between the tongue and the floor of the mouth is too tight or too short. This may cause feeding problems for the baby and/or nipple pain for a breastfeeding mother.

Study characteristics: Five randomised controlled trials enrolling 302 infants met the inclusion criteria.

Key results: In an infant with tongue-tie and feeding difficulties, surgical release of the tongue-tie does not consistently improve infant feeding but is likely to improve maternal nipple pain. Further research is needed to clarify and confirm this effect.

Quality of evidence: The quality of the evidence is very low to moderate because overall only a small number of studies have looked at this condition, the total number of babies included in these studies was low and some studies could have been better designed.

SUMMARY OF FINDINGS FOR THE MAIN COMPARISON [Explanation]

Undefined

Patient or population: tongue-tie in newborn infants
Setting: maternity hospitals
Intervention: frenotomy
Comparison: no frenotomy or sham procedure

Outcomes	Illustrative comparative risks (mean and SD)		Relative effect (95% CI)	Number of participants (studies)	Quality of the evidence (GRADE)	Comments
	Risk with no frenotomy or sham procedure	Risk with frenotomy				
Infant breastfeeding assessed by validated scale - IBFAT scores following procedure	Mean IBFAT scores following procedure in the control group was 8.1 (SD 0.9)	Mean IBFAT scores following procedure in the frenotomy group was 11.6 (SD 0.8)	Mean difference is 3.50 (3.06 to 3.94)	58 (1 RCT)	⊕⊕◯◯ LOW[a,b]	IBFAT score is based on a 12-point scale
Infant breastfeeding assessed by validated scale - LATCH scores following procedure	Mean LATCH scores following procedure in the control group was 6.8 to 8.5 (SD < 1.9)	Mean LATCH scores following procedure in the frenotomy group was 6.8 to 8.4 (SD < 2)	Mean difference is -0.07 (-0.63 to 0.48)	155 (2 RCTs)	⊕⊕◯◯ LOW[a,b]	LATCH score is based on a 10-point scale
Maternal nipple pain assessed by a validated pain scale - visual analogue pain scale	Mean visual analogue pain scale scores following procedure in the control group was 2.9 to 5.5 (SD < 2.6)	Mean IBFAT scores following procedure in the frenotomy group was 1.6 to 5.3 (SD < 2.4)	Mean difference is -0.74 (-1.35 to -0.13)	183 (3 RCTs)	⊕⊕◯◯ LOW[a,b]	Visual analogue pain scale score is based on a 10-point scale
Maternal nipple pain assessed by a validated pain scale - SF-MPQ pain scale	Mean SF-MPQ scores following procedure in the control group was 13.5 (SD 1.5)	Mean IBFAT scores following procedure in the frenotomy group was 4.9 (SD 1.5)	Mean difference is -8.60 (-9.37 to -7.83)	58 (1 RCT)	⊕⊕◯◯ LOW[a,b]	SF-MPQ score is based on a 50-point scale

*__The risk in the intervention group__ (and its 95% confidence interval) is based on assumed risk in the comparison group and the __relative effect__ of the intervention (and its 95% CI)

CI: confidence interval; OR: odds ratio; RR: risk ratio

GRADE Working Group grades of evidence
High quality: We are very confident that the true effect lies close to the estimate of effect
Moderate quality: We are moderately confident in the effect estimate: The true effect is likely to be close to the estimate of effect but may be substantially different
Low quality: Our confidence in the effect estimate is limited: The true effect may be substantially different from the estimate of effect
Very low quality: We have very little confidence in the effect estimate: The true effect is likely to be substantially different from the estimate of effect

[a] Imprecision (small total participant population)
[b] Risk of bias (incomplete blinding)

BACKGROUND

Description of the condition

The lingual frenulum is a fold of mucous membrane that extends from the floor of the mouth to the midline of the underside of the tongue. It helps to stabilise the base of the tongue and does not normally interfere with tongue tip movement (Marchesan 2005). Tongue-tie (ankyloglossia) is a condition in which the lingual frenulum has an anterior attachment near the tip of the tongue and may be unusually short, tight and thick (Jackson 2012). This causes virtual adhesion of the tongue tip to the floor of the mouth and can result in restricted tongue tip movement (Marchesan 2005). The exact cause of 'tongue-tie' (ankyloglossia) is not known. Genetics may play a role, as the condition tends to run in some families (Coryllos 2004). Prevalence is about 4% to 11% among newborns (Hogan 2005; Messner 2000a; Messner 2000b; Ricke 2005).

Tongue-tie has been cited as a cause of poor breastfeeding because the infant is unable to attach or stay latched on, and because maternal nipple pain may result (Coryllos 2004; Hogan 2005). In older children and adults, tongue-tie has been implicated as a cause of speech delay, abnormal dentition, poor oral hygiene and inability to play wind instruments (Krol 2007). As an infant breastfeeds, the tongue moves with peristalsis over maternal lactiferous sinuses and extracts milk. When the infant's tongue movement is restricted, as is the case with severe tongue-tie, reduced movement may affect milk extraction, and friction may be present between the tongue or gums and the nipple, causing damage to the nipple and maternal pain (Coryllos 2004; Hogan 2005). References to tongue-tie causing speech problems date back to Aristotle in the third century BC (Obladen 2010). The association between breastfeeding difficulty and tongue-tie has been recognised for at least 500 years (Obladen 2010). In recent years, with recognition and encouragement of exclusive breastfeeding as the optimal primary mode of infant feeding, the justification for frenotomy has shifted from improving speech problems to improving breastfeeding (Obladen 2010), re-igniting the historical debate as to the role of tongue-tie in breastfeeding difficulties (Kumar 2012).

The diagnosis of tongue-tie depends on an assessment of the structure and function of the lingual frenulum. Diagnostic classification systems vary from simple visual inspection and/or palpation of the frenulum to a more complex multi-scale classification system such as the Hazelbaker Assessment Tool for Lingual Frenulum Function (ATLFF) (Hazelbaker 1993). The ATLFF is a highly reliable screening tool (Amir 2006) that was designed to be used in assessment of infants younger than three months of age. It assesses the function and appearance of the frenulum. A score of 14 indicates normal function, between 11 and 14 is acceptable and less than 11 indicates significant tongue-tie that requires frenotomy. An appearance scale includes values to 10, and lower scores indicate tongue-tie. A score lower than eight is suggestive of tongue-tie, but surgery is not recommended unless a functional problem is noted.

Description of the intervention

Frenotomy and frenuloplasty are the two main surgical procedures used in the treatment of infants with tongue-tie (Lalakea 2002). Frenotomy, or clipping of the frenulum, is the procedure of choice in infants because it is relatively quick and easy to perform. The infant is swaddled and is placed supine on the examining table, while an assistant supports the head and neck. The clinician/surgeon elevates the tongue and exposes the frenulum, which is then incised with sharp, straight, blunt-ended scissors (Berry 2012; Hogan 2005). Some operators describe crushing the frenulum before incision. Direct pressure is then applied to the frenulum with a piece of gauze. Bleeding is reportedly scant and is controlled by the pressure (Lalakea 2002). The incision usually is not sutured, and the infant most often recovers quickly from the procedure and is able to feed directly afterwards. In infants, frenotomy is usually performed without analgesia or anaesthetic (Lalakea 2002). The use of a laser to perform the frenotomy is becoming more frequent (Kotlow 2011).

Frenuloplasty, an operation that lengthens the frenulum, is the preferred procedure for patients over one year of age (Lalakea 2002). This intervention will not be included in this review.

How the intervention might work

Surgical release of the tongue-tie through frenotomy may correct the restriction to infant tongue movement during feeding to allow more effective breastfeeding and less maternal nipple pain resulting from decreased friction between the infant's lower gum/tongue and the nipple (Kumar 2012).

Why it is important to do this review

Diagnosis and management of tongue-tie remain controversial. It is uncertain whether ankyloglossia is a congenital oral anomaly requiring treatment or a normal variant. One survey (Messner 2000b) found that most lactation consultants believe tongue-tie to be a frequent cause of infant breastfeeding difficulties that could be solved by frenotomy. In marked contrast, 90% of paediatricians and 70% of otolaryngologists believe that tongue-tie never, or rarely, causes a feeding problem (Messner 2000a). However, medical organisations such as the American Academy of Pediatrics (Coryllos 2004) and the National Institute for Health and Care Excellence (NICE 2005) now acknowledge that tongue-tie, or ankyloglossia, is a significant clinical entity that should be treated as early as possible to minimise breastfeeding problems. Given that breastfeeding benefits both infants and mothers, it is important

for the clinician to address any condition that may impair breastfeeding (Edmunds 2011).

OBJECTIVES

To determine whether frenotomy is safe and effective in improving ability to feed orally among infants younger than three months of age with tongue-tie (and problems feeding).

Subgroup analysis

- Severity of tongue-tie before frenotomy as measured by a validated tool (e.g. ATLFF (scores < 11; scores ≥ 11)) (Hazelbaker 1993).
- Gestational age at birth (< 37 weeks' gestation; 37 weeks' gestation and above).
- Method of feeding (breast or bottle).
- Age at frenotomy (≤ 10 days of age; > 10 days to three months of age).
- Severity of feeding difficulty (infants with feeding difficulty affecting weight gain (as assessed by infant's not regaining birth weight by day 14 or falling off centiles); infants with symptomatic feeding difficulty but thriving (greater than birth weight by day 14 and tracking centiles).

METHODS

Criteria for considering studies for this review

Types of studies

Randomised or quasi-randomised controlled trials or cluster-randomised trials.

Types of participants

Infants three months of age or younger with a diagnosis of tongue-tie who are orally feeding. To be included, another problem must be present that could be related to the tongue-tie, specifically, infant feeding problems or maternal nipple pain in a breastfeeding mother. We planned to exclude patients with other coexisting oral pathology that might affect oral feeding, for example, cleft palate.

Types of interventions

- Frenotomy versus no frenotomy. Lactation consultant interventions were accepted if provided to both groups.
- Frenotomy versus sham procedure. Lactation consultant interventions were accepted if provided to both groups.

Types of outcome measures

Primary outcomes

- Infant feeding assessed within 48 hours, within two to seven days and after seven days following the procedure with the use of a validated scale such as the LATCH score (Jensen 1994) or the Infant Breastfeeding Assessment Tool (IBFAT) (Mathews 1988). The LATCH is a scoring system that assesses latch, swallowing, maternal nipple, maternal comfort and assistance the mother needs to position the infant. Each area gets a score between 0 and 2 with a total possible score of 10. The IBFAT assesses readiness to feed, rooting, fixing (latching on) and sucking. Each item is scored between 0 and 3, and a score of 10 to 12 represents successful breastfeeding. Bottle-feeding will be assessed within 48 hours, within two to seven days and after seven days following the procedure, subjectively, by reports of more efficient sucking and less drooling

Secondary outcomes

- Maternal nipple pain assessed within 48 hours, within two to seven days and after seven days following frenotomy by a validated pain scale (e.g. Short-Form McGill Pain Questionnaire (SF-MPQ)) (Melzack 1975)
- Qualitative assessment of infant feeding by parental survey performed within 48 hours of the procedure
- Duration of breastfeeding (days)
- Cessation of breastfeeding as assessed by maternal report within four weeks of the procedure
- Infant pain as assessed by a validated pain scale (e.g. Modified Behavioral Pain Scale (MBPS) (Taddio 1995), Neonatal Infant Pain Scale (NIPS) (Lawrence 1993), CRIES pain scale (cries, requires oxygen, shows increased vital signs and expression and is sleepless) (Krechel 1995)) before, during and up to one hour post frenotomy
- Excessive bleeding at the time or within 24 hours of frenotomy (as determined by study investigators)
- Infection at the site of frenotomy requiring treatment with antibiotics within seven days of the procedure
- Damage to the tongue or submandibular ducts noted within seven days of the procedure (as determined by study investigators)

Search methods for identification of studies

Electronic searches

Two review authors (JO'S, JF) independently performed electronic database searches, including electronic searches of the Cochrane Central Register of Controlled Trials (CENTRAL; 2016, Issue 1),

MEDLINE (1966 to January 2016), Embase (1980 to January 2016) and the Cumulative Index to Nursing and Allied Health Literature (CINAHL; 1982 to January 2016), and searched previous reviews including cross-references, expert informants and journal handsearching. We searched MEDLINE, Embase and CINAHL for relevant articles using the following search terms: Infant AND Tongue Tie, Infant OR Newborn OR neonate (explode) [MeSH heading] AND Tongue Tie (explode) [MeSH heading] OR ankyloglossia. and Frenotomy [MeSH heading] OR Frenulotomy OR Frenuloplasy [MeSH heading]. We applied no language restrictions. We also searched clinical trial registries for current and recently completed trials (Australia and New Zealand Clinical Trials Register (ANZCTR); clinicaltrials.gov; controlled-trials.com; who.int/ictrp; and Oxford Database of Perinatal Trials).

Searching other resources

The search strategy included communication with expert informants and searches of bibliographies of reviews and trials for references to other trials, as well as searches of previous reviews including cross-references, abstracts and conferences and symposia proceedings of the Perinatal Society of Australia and New Zealand and the Pediatric Academic Societies (American Pediatric Society, Society for Pediatric Research and European Society for Pediatric Research) from 1990 to 2015. We planned to contact the corresponding investigator of any unpublished trials to request information. We considered unpublished studies and studies reported only as abstracts as eligible for review if final trial data were available. We intended to contact the corresponding authors of identified randomised controlled trials (RCTs) to ask for additional information about their studies if we required further data.

Data collection and analysis

We used the standard methods of Cochrane as documented in the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011) and the methods of the Cochrane Neonatal Review Group (CNRG).

Selection of studies

Review authors independently assessed for inclusion all potential studies identified by the search strategy. We resolved disagreements through discussion.

Specifically, we:

- merged search results by using reference management software and removed duplicate records of the same report;
- examined titles and abstracts to remove irrelevant reports;
- retrieved full text of potentially relevant reports;
- linked together multiple reports of the same study;
- examined full-text reports to assess for compliance of studies meeting eligibility criteria;
- corresponded with investigators, when appropriate, to clarify study eligibility;
- at all stages noted reasons for inclusion and exclusion of articles; and
- made final decisions on study inclusion and proceeded to data collection.

Data extraction and management

Review authors independently extracted data from full-text articles using a specifically designed spreadsheet to manage the information. We resolved discrepancies through discussion; if required, we intended to consult a review arbiter. We entered data into Review Manager software 5.3 (RevMan 2014) and checked them for accuracy.

Assessment of risk of bias in included studies

We used the standardised review methods of the CNRG (http://neonatal.cochrane.org/en/index.html) to assess the methodological quality of included studies. Review authors independently assessed study quality and risk of bias using the following criteria, as documented in the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011).

- Random sequence generation: Was the allocation sequence adequately generated?
- Allocation concealment: Was allocation adequately concealed?
- Blinding of participants and personnel for each main outcome or class of outcomes: Was knowledge of the allocated intervention adequately prevented during the study?
- Blinding of outcome assessors: Were the outcome assessors blinded?
- Incomplete outcome data for each main outcome or class of outcomes: Were incomplete data adequately addressed?
- Selective outcome reporting: Are reports of the study free of the suggestion of selective outcome reporting? We tried to locate protocols to assess outcome reporting bias.
- Other sources of bias: Was the study apparently free of other problems that could put it at high risk of bias? We gave particular attention to completeness of follow-up of all randomly assigned infants and to the length of follow-up studies to identify whether any benefits claimed were robust.

We intended to request additional information and clarification of published data from the authors of individual trials if required. We assessed each trial for risk of bias based on the criteria listed above and marked each as:

- 'low' risk of bias;
- 'unclear' risk of bias; or
- 'high' risk of bias.

We judged each criterion as being at 'low risk' of bias, 'high risk' of bias or 'unclear' risk of bias (for lack of information or uncertainty over the potential for bias).

Measures of treatment effect

We analysed the results of included studies using the statistical package Review Manager software 5.3 (RevMan 2014).

We used the standard method of the CNRG, applying a fixed-effect model for meta-analysis. In assessing treatment effects for dichotomous data, we reported the risk ratio (RR) and the risk difference (RD), along with 95% confidence intervals, for categorical outcomes. If the RD was statistically significant, we calculated the number needed to treat for an additional beneficial outcome (NNTB) and the number needed to treat for an additional harmful outcome (NNTH) (1/RD). For outcomes measured on a continuous scale, we used the weighted mean difference, along with 95% confidence intervals.

Included studies reported outcomes of change in infant feeding ability and in maternal pain using different validated scales. For infant feeding, researchers presented LATCH scores (a 10-point scale) and IBFAT scores (a 12-point scale). Included studies reported maternal pain assessed by the 10-point visual analogue pain scale and the 50-point SF-MPQ. As these different scales rely on very different units of reporting and show subtle differences, we did not combine scores for analysis, and we presented results in subgroups according to the different scales used.

Unit of analysis issues

We combined randomised trials in a single meta-analysis using the generic inverse variance method.

Dealing with missing data

We planned to contact the authors of all published studies if we required clarification or additional information. We planned to describe the number of participants with missing data in the Results section and in the Characteristics of included studies table. We presented results only for available participants. We intended to discuss the implications of missing data in the Discussion section of the review.

Assessment of heterogeneity

We used RevMan 5.3 (RevMan 2014) to assess the heterogeneity of treatment effects between trials. We used the two formal statistical approaches described below.

- Chi^2 test for homogeneity: We calculated whether statistical heterogeneity is present by using the Chi^2 test for homogeneity ($P < 0.1$). Because this test has low power when the number of studies included in the meta-analysis is small, we set the probability at the 10% level of significance (Higgins 2011).
- I^2 statistic, to ensure that pooling of data was valid: We quantified the impact of statistical heterogeneity by using I^2 statistics available in RevMan 2014, which describes the percentage of total variation across studies due to heterogeneity rather than to sampling error. We graded the degree of heterogeneity as follows: 0% to 30%: potentially trivial (not important) heterogeneity; 31% to 50%: low heterogeneity; 51% to 75%: moderate heterogeneity; and 76% to 100%: high heterogeneity. Had we found evidence of apparent or statistical heterogeneity, we planned to assess the source of heterogeneity by performing sensitivity and post hoc subgroup analyses to look for sources of bias or methodological differences between heterogeneous trials (e.g. differences in study quality, participants, intervention regimens, outcome assessments).

Assessment of reporting biases

We tried to obtain the study protocols of all included studies to compare outcomes reported in the protocol versus those reported in the findings for each of the included studies. We intended to investigate reporting and publication bias by examining the degree of asymmetry of a funnel plot if we identified 10 or more trials. When we suspected reporting bias (see selective reporting bias above), we intended to contact study authors to ask them to provide missing outcome data. When this was not possible and we suspected that missing data might introduce serious bias, we intended to explore the impact of including such studies in the overall assessment of results by performing a sensitivity analysis.

Data synthesis

We performed statistical analyses according to the recommendations of the CNRG (http://neonatal.cochrane.org/en/index.html). We analysed all randomly assigned infants on an intention-to-treat (ITT) basis. We analysed treatment effects in individual trials and used a fixed-effect model for meta-analysis in the first instance to combine the data. When substantial heterogeneity existed, we examined the potential cause of heterogeneity by performing subgroup and sensitivity analyses. When we judged the meta-analysis to be inappropriate, we analysed and interpreted individual trials separately. For estimates of typical risk ratio and risk difference, we used the Mantel-Haenszel method. For measured quantities, we used the inverse variance method.

Quality of evidence

We used the Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach, as outlined in the GRADE Handbook (Schünemann 2013), to assess the quality of evidence for infant breastfeeding when assessed by a validated scale and maternal nipple pain.

Two review authors independently assessed the quality of the evidence for each of the outcomes above. We considered evidence from RCTs as high quality but downgraded the evidence one level for serious (or two levels for very serious) limitations on the basis of the following: design (risk of bias), consistency across studies, directness of the evidence, precision of estimates and presence of

publication bias. We used the GRADEpro Guideline Development Tool to create Summary of findings for the main comparison to report the quality of the evidence.
The GRADE approach results in an assessment of the quality of a body of evidence according to one of four grades.

- High: We are very confident that the true effect lies close to that the estimate of effect.
- Moderate: We are moderately confident in the effect estimate: The true effect is likely to be close to the estimate of effect but may be substantially different.
- Low: Our confidence in the effect estimate is limited: The true effect may be substantially different from the estimate of effect.
- Very low: We have very little confidence in the effect estimate: The true effect is likely to be substantially different from the estimate of effect

Subgroup analysis and investigation of heterogeneity

We planned to carry out the following subgroup analyses.

- Severity of tongue-tie as measured by a validated tool (e.g. ATLFF (scores < 11; scores ≥ 11)) (Hazelbaker 1993).
- Gestational age at birth (< 37 weeks' gestation; 37 weeks' gestation and above).
- Method of feeding (breast or bottle).
- Age at frenotomy (≤ 10 days of age; > 10 days to three months of age).
- Severity of feeding difficulty (infants with feeding difficulty affecting weight gain (assessed by infant's not regaining birth weight by day 14 or dropping off centiles by three months); infants with feeding difficulty but normal weight gain).

Sensitivity analysis

We intended to explore methodological heterogeneity through the use of sensitivity analysis. We classified studies as having low risk of bias if they had adequate sequence generation and allocation concealment and reported losses less than 10% on ITT analysis.

RESULTS

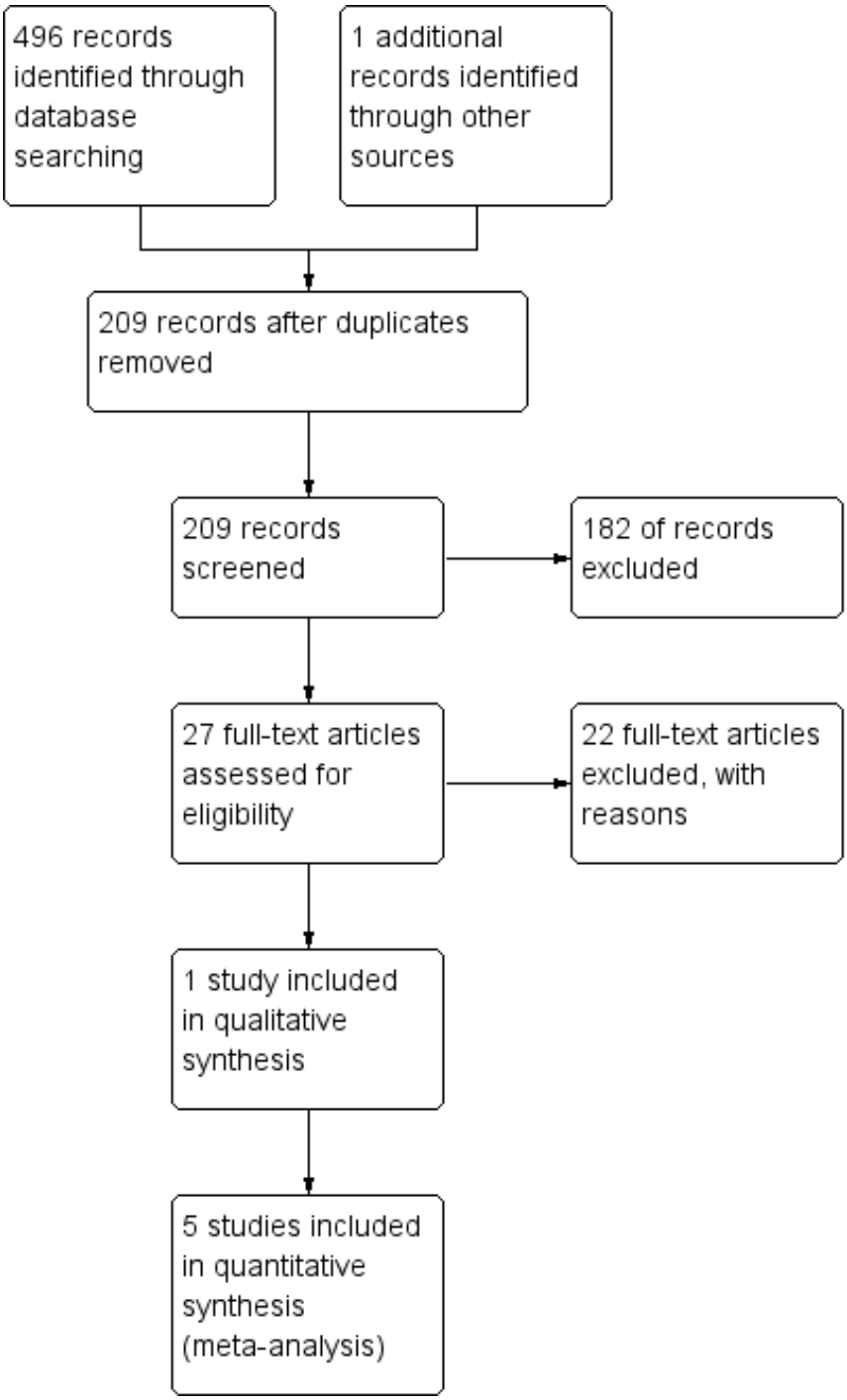
Description of studies

See Characteristics of included studies and Characteristics of excluded studies.

Results of the search

We present a summary of our search in Figure 1. We encountered no disagreement between assessors (JO'S, JF, DT) regarding inclusion or exclusion of studies, quality assessment or data extraction. We pooled available data and analysed them as listed below.

Figure 1. Study flow diagram.



Included studies

We included five small trials in this review (N = 302).

- Berry 2012 is a single-centre study performed in the United Kingdom.
  - Objective: to investigate if an immediate and sustained improvement in breastfeeding occurs following frenotomy.
  - Population: infants younger than four months of age with tongue-tie and breastfeeding difficulties.
  - Intervention: infants randomised to frenotomy or no frenotomy.
  - Outcomes: subjective and objective feeding assessment and maternal pain scores.

- Buryk 2011 is a single-centre study performed in America.
  - Objective: to investigate if frenotomy decreased maternal pain, improved breastfeeding scores and led to breastfeeding over a longer period.
  - Population: infants younger than 30 days of age with breastfeeding difficulties whose mothers experienced nipple pain.
  - Intervention: infants randomised to frenotomy or sham procedure.
  - Outcomes: change in maternal pain scores and infant feeding scores.

- Dollberg 2006 is a single-centre study performed in Israel.
  - Objective: to investigate if frenotomy in infants with ankyloglossia improves nipple pain and/or latching on during attempts to breastfeed.
  - Population: breastfeeding infants younger than 21 days of age whose mothers were experiencing nipple pain.
  - Intervention: infants randomised to frenotomy followed by a sham procedure or a sham procedure followed by frenotomy.
  - Outcomes: change in maternal pain scores and infant latch scores.

- Emond 2013 is a single-centre study performed in the United Kingdom.
  - Objective: to investigate if immediate frenotomy was superior to breastfeeding support.
  - Population: breastfeeding infants younger than two weeks of age with mild to moderate tongue-tie and breastfeeding difficulties.
  - Intervention: infants randomised to frenotomy or standard care.
  - Outcomes: change in breastfeeding scores and maternal pain scores.

- Hogan 2005 is a single-centre study performed in the United Kingdom.
  - Objective: to investigate if frenotomy was superior to standard care in infants with tongue-tie and feeding difficulties.
  - Population: bottle-feeding and breastfeeding infants with tongue-tie and feeding difficulty.
  - Intervention: infants randomised to intensive lactation support or frenotomy.
  - Outcomes: subjective improvement in feeding.

Excluded studies

See Characteristics of excluded studies. We excluded two studies (Ngerncham 2013; Yousefi 2015) - the first because it was a cross-sectional study with no control group, the second because it compared frenotomy versus frenuloplasty in children up to 12 years of age.

Risk of bias in included studies

We included in the analysis randomised controlled trials that compared frenotomy versus no frenotomy or frenotomy versus sham procedure in newborn infants. Overall, the five included trials had small study populations and a high incidence of methodological shortcomings. We discuss below specific methodological issues regarding these studies. See the risk of bias graph in Figure 2 and summary in Figure 3.

Figure 2. Risk of bias graph: review authors' judgements about each risk of bias item presented as percentages across all included studies.

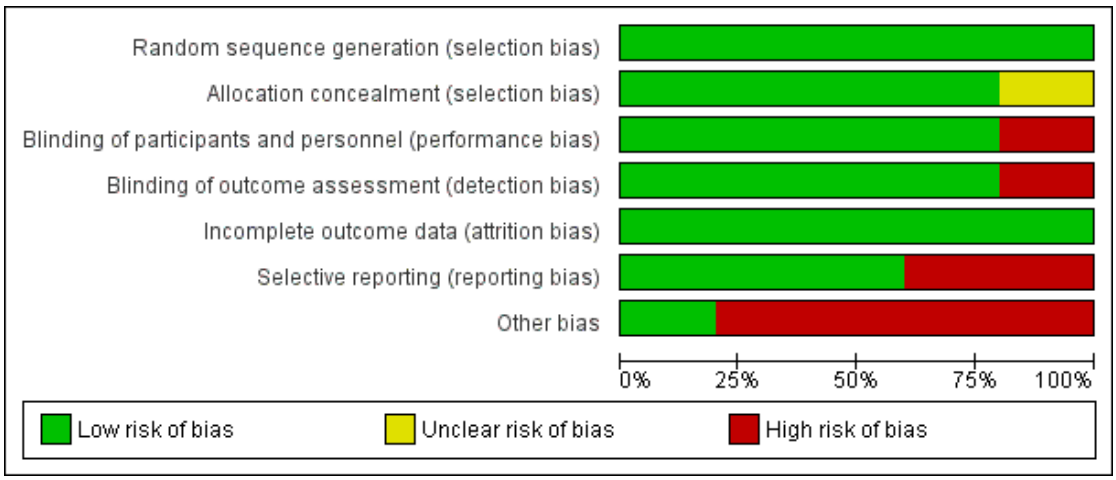
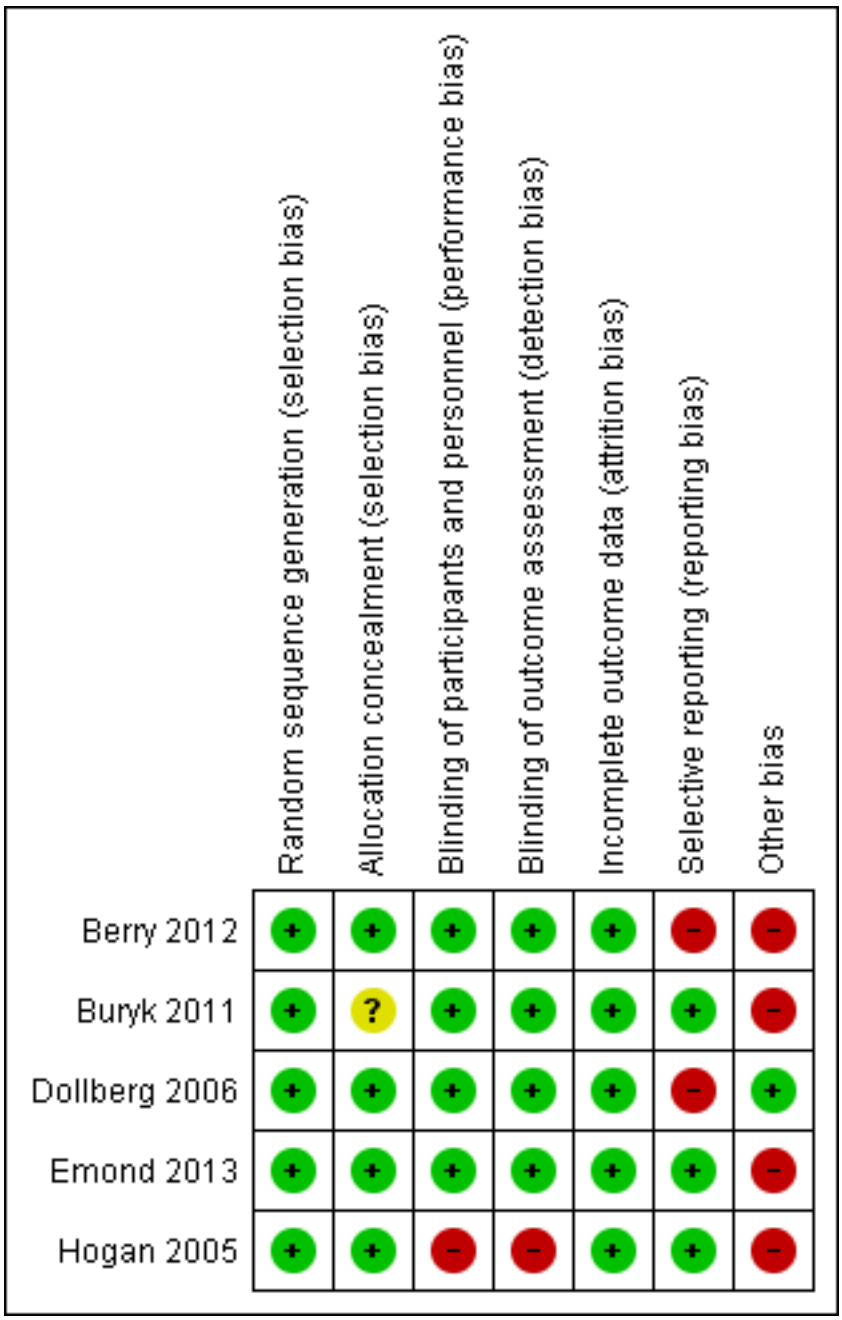


Figure 3. Risk of bias summary: review authors' judgements about each risk of bias item for each included study.



Allocation

Random sequence generation

All five studies reported the method of randomisation. Berry 2012, Buryk 2011, Dollberg 2006 and Hogan 2005 used computer-generated randomisation, and Emond 2013 used independent telephone-based randomisation.

Allocation concealment

Four studies described the methods used to conceal allocation. Berry 2012 and Hogan 2005 used sealed envelopes created by a person not involved in the trial, and Dollberg 2006 used sealed envelopes and specified that the envelopes were opaque but did not state who created them. Emond 2013 used a randomisation code that was kept by an independent body. Buryk 2011 did not state how investigators concealed allocation.

Blinding

Blinding of participants and research or clinical personnel

All five studies reported this. Hogan 2005 did not blind participants nor personnel. Buryk 2011 performed single-blinding (mothers only, not treating personnel), Emond 2013 also used single-blinding (researchers blinded, mothers not blinded) and Berry 2012 and Dollberg 2006 blinded both participants and personnel.

Blinding of outcome assessment

All five studies reported this. Berry 2012, Buryk 2011 and Dollberg 2006 blinded the assessor. Emond 2013 blinded the assessor of the primary outcome and assessors of some secondary outcomes but did not blind the mother and did not blind the assessors of other secondary outcomes. Hogan 2005 was an unblinded study.

Incomplete outcome data

Exclusions after randomisation

Berry 2012 had three postrandomisation exclusions (5% of randomised participants), and Dollberg 2006 had one postrandomisation exclusion (4% of randomised participants). Both studies reported that exclusions were due to failure of blinding. Emond 2013 had one postrandomisation exclusion (< 1% of randomised participants) resulting from loss to follow-up. Buryk 2011 and Hogan 2005 had no postrandomisation exclusions.

Selective reporting

Two studies selectively reported outcomes. Berry 2012 reported that infant latch onto the breast objectively improved but did not provide data; Berry 2012 also reported on breastfeeding success at three months but did not report results by study group. Buryk 2011 reported no differences between groups in longer-term breastfeeding rates but did not present results of the two groups separately. Dollberg 2006 reported changes in pain scores before and after the intervention in the intervention arm only and on request produced scores for the control arm.

Other potential sources of bias

All five studies offered and provided frenotomy to control infants, suggesting lack of equipoise. Two studies - Berry 2012 and Dollberg 2006 - performed frenotomy on all participants as part of the study protocol. Buryk 2011, Emond 2013 and Hogan 2005 offered frenotomy to controls and had uptakes of 77% to 97%.

Effects of interventions

See: **Summary of findings for the main comparison** Frenotomy compared with no frenotomy or sham procedure in infants with tongue-tie and feeding difficulties

Frenotomy versus no frenotomy or sham procedure

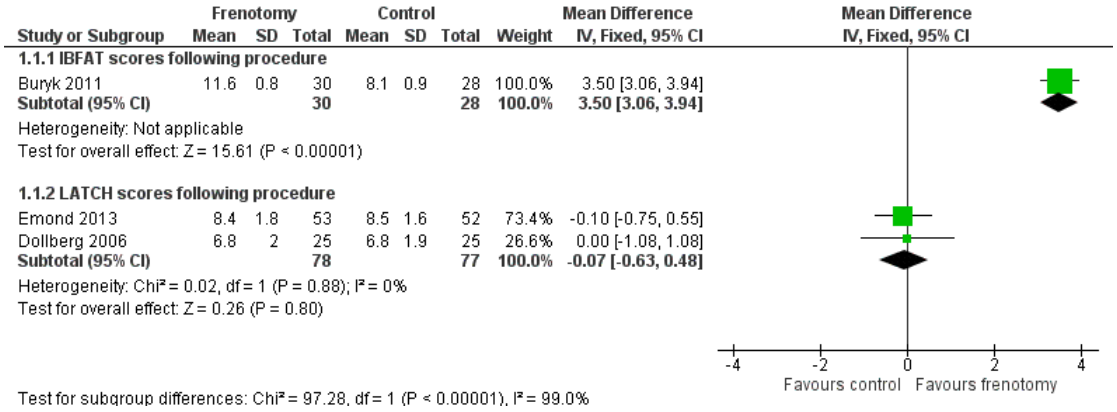
Five studies compared frenotomy versus no frenotomy (Berry 2012; Emond 2013; Hogan 2005) or frenotomy versus sham procedure (Buryk 2011; Dollberg 2006). We summarise the main findings in Summary of findings for the main comparison.

Primary outcomes

Infant feeding assessed with a validated tool (Analysis 1.1)

Four trials provided outcome data for this comparison (Berry 2012; Buryk 2011; Dollberg 2006; Emond 2013). Three studies reported improvement in infant breastfeeding assessed with a validated scale (Buryk 2011; Dollberg 2006; Emond 2013) based on two different measurement scales (Analysis 1.1; Figure 4).

Figure 4. Forest plot of comparison: 1 Frenotomy versus no frenotomy or sham procedure, outcome: 1.1 Infant breastfeeding assessed by a validated scale.



- IBFAT score (Buryk 2011): MD 3.50 on a 12-point scale, 95% CI 3.06 to 3.94; 58 participants (Analysis 1.1.1).

- LATCH score (Dollberg 2006; Emond 2013): MD -0.07 on a 10-point scale, 95% CI -0.63 to 0.48; 155 participants (heterogeneity: Chi^2 = 0.02, df = 1, P = 0.88; I^2 = 0%) (Analysis 1.1.2).

A fourth trial (Berry 2012) also reported that infants in the intervention group had higher LATCH scores after the intervention but provided no data.
We did not combine LATCH and IBFAT scores for meta-analysis, as studies had a high degree of heterogeneity and scales used different units of measure.

Subgroup analyses

Severity of tongue-tie

One trial included only infants with severe tongue-tie (Buryk 2011) and reported improvement in infant breastfeeding (increase in IBFAT scores) in the frenotomy group compared with the control group: MD 3.50 on a 12-point scale, 95% CI 3.06 to 3.94; 58 participants.
One trial included only infants with moderate tongue-tie (Emond 2013) and found no objective improvement in feeding scores: MD -0.10 on a 10-point scale, 95% CI -0.75 to 0.55; 105 participants.

Timing of feeding assessment

Two trials reported on infant breastfeeding within 48 hours of the intervention (Buryk 2011; Dollberg 2006).

- IBFAT score (Buryk 2011): MD 3.50 on a 12-point scale, 95% CI 3.06 to 3.94; 58 participants.

- LATCH score (Dollberg 2006): MD 0.0 on a 10-point scale, 95% CI -1.08 to 1.08; 50 participants.

One trial (Emond 2013) reported no difference in LATCH scores assessed between two and seven days after the intervention: MD -0.10 on a 10-point scale, 95% CI -0.75 to 0.55; 105 participants (Analysis 1.2).
Subgroup analysis by *gestational age* or by *severity of feeding difficulty* or*method of feeding* was not possible owing to lack of data.

Secondary outcomes

Effect on maternal nipple pain (Analysis 1.3)

Four trials reported this outcome (Berry 2012; Buryk 2011; Dollberg 2006; Emond 2013) using two different scales.

- Berry 2012, Dollberg 2006 and Emond 2013 assessed this outcome after the first breastfeed using a 10-point visual analogue pain scale. Meta-analysis showed a significant reduction in maternal nipple pain in the frenotomy group compared with the control group: MD -0.74, 95% CI -1.35 to -0.13 (heterogeneity: Chi^2 = 1.85, df = 2, P = 0.40; I^2 = 0%) (Analysis 1.3.1).

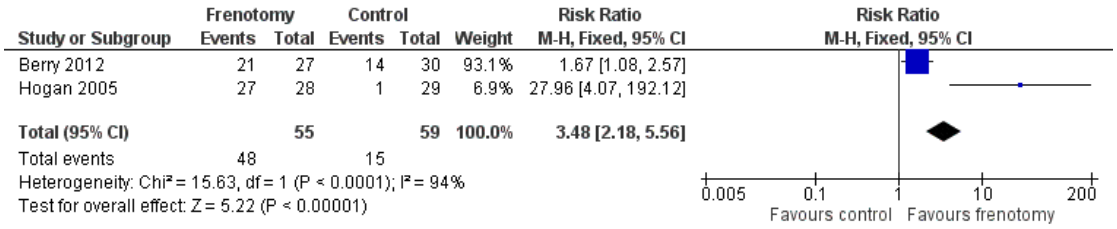
- Buryk 2011 used the 50-point SF-MPQ Pain Assessment Tool after five days of feeding and found a significant reduction in the frenotomy group compared with the control group: MD -8.60, 95% CI -9.37 to -7.83 (Analysis 1.3.2).

We did not combine visual analogue pain scale and SF-MPQ scores for meta-analysis because studies had a high degree of heterogeneity and scales used different units of measure.

Qualitative assessment of infant feeding by parental survey performed within 48 hours of the procedure (Analysis 1.4)

Two trials (Berry 2012; Hogan 2005) reported high rates of improvement in breastfeeding following frenotomy compared with control (typical risk ratio (RR) 3.48, 95% CI 2.18 to 5.56). The meta-analysis contained high levels of heterogeneity ($Chi^2 = 15.63$, df = 1, $P < 0.0001$; $I^2 = 94\%$) (Figure 5).

Figure 5. Forest plot of comparison: 1 Frenotomy versus no frenotomy or sham procedure, outcome: 1.6 Qualitative assessment of infant feeding by parental survey performed within 48 hours of the procedure.



Hogan 2005 reported that all intervention infants (n = 8) versus no control infants (n = 9) had improved bottle-feeding.

Emond 2013 obtained qualitative data from a subgroup of the study population and found that most parents reported improvement in infant feeding directly after the intervention and in the days that followed, but investigators did not state when they performed the survey.

Duration of breastfeeding or cessation of breastfeeding as assessed by maternal report within four weeks of the procedure

No trial reported the duration of breastfeeding or cessation of breastfeeding as assessed by maternal report within four weeks. However, four studies reported on rates of breastfeeding at specific time points. Berry 2012 reported that 58/59 were breastfeeding at three months but did not report this by treatment group. Buryk 2011 reported that 36/58 were breastfeeding at two months, 23/58 at six months and 14/58 at 12 months. Study authors also did not report by treatment group. Emond 2013 reported that at eight weeks, 43/52 in the intervention group and 40/50 in the control group were at least partially breastfed. Hogan 2005 reported that 12/20 in the intervention group and 10/20 in the control group were breastfed for at least four months.

Infant pain assessed by a validated pain scale before, during and up to one hour post frenotomy

None of the included studies reported this outcome.

Excessive bleeding at the time or within 24 hours of frenotomy (Analysis 1.5)

No infants in any of the five trials experienced excessive bleeding following the intervention (RR not estimable; RD 0.00, 95% CI -0.03 to 0.03; 302 participants).

Infection at the site of frenotomy requiring treatment with antibiotics within seven days of the procedure (Analysis 1.6)

No infants in any of the five trials experienced infection at the intervention site requiring antibiotics (RR not estimable; RD 0.00, 95% CI -0.03 to 0.03; 302 participants).

Damage to the tongue or submandibular ducts noted within seven days of the procedure (Analysis 1.7)

No infants in any of the five trials experienced damage to the tongue or submandibular ducts (RR not estimable; RD 0.00, 95% CI -0.03 to 0.03; 302 participants).

DISCUSSION

Summary of main results

We included in this review five trials with a total sample size of 302 infants. Only one small trial reported that frenotomy objectively

improved infant breastfeeding (Buryk 2011). Pooled analysis of two other trials (N = 155) showed no objective change following frenotomy (Dollberg 2006; Emond 2013) but showed consistently reduced maternal nipple pain.

Studies to date have not answered the clinically relevant question of whether frenotomy in tongue-tied infants with feeding difficulties results in longer-term breastfeeding success and resolution of maternal pain. No studies reported duration of breastfeeding by group, and all had very high contamination rates in the control group, making longer-term outcomes meaningless.

Overall completeness and applicability of evidence

All studies were performed at maternity hospitals in the United Kingdom, America and Israel. All centres that carried out the studies had lactation support and frenotomy readily available. The total number of infants studied to date is 302. This number is too small to confirm the efficacy and safety of frenotomy, even though no study reported any significant adverse effects. Populations studied to date were healthy term infants with moderate or severe tongue-tie. All primary outcomes were short-term, many assessed after just one feed following intervention. Some studies followed up infants for several months after the intervention, but as the contamination rate within the control group was very high, longer-term results are meaningless. Studies to date have not answered the clinically relevant question of whether frenotomy in tongue-tied infants results in longer-term breastfeeding success and resolution of maternal pain.

The potential to improve infant breastfeeding while reducing maternal nipple pain may be greatest when an infant is given the diagnosis of severe tongue-tie. However, only one trial has studied frenotomy in this population. Again, only one study has examined frenotomy in infants given the diagnosis of moderate tongue-tie and found that frenotomy did not have an objective effect on infant feeding nor on maternal nipple pain but was subjectively effective.

Quality of the evidence

We assessed the quality of the evidence using the GRADE method and classified evidence for major outcomes as low quality on the basis of small sample sizes (both in individual studies and in combined studies), inconsistent blinding and high risk of bias. The included studies had low rates of participant drop-out. Most study authors responded when we requested further information. We noted high degrees of heterogeneity between studies and observed that investigators used different diagnostic tools to diagnose the condition, reported differing degrees of tongue-tie severity and used different scales to assess outcomes. The greatest weakness among studies to date is that investigators offered frenotomy to all controls and provided the procedure for a large majority of them, which strongly implies lack of equipoise about effectiveness of the intervention during the study and reduces the quality of results.

Potential biases in the review process

We used the standard methods of the Cochrane Neonatal Review Group in conducting this systematic review. Our inclusive search strategy would have included all relevant studies.

Agreements and disagreements with other studies or reviews

Several other published reviews have examined effects of frenotomy for tongue-tie in young infants (Algar 2009; Bowley 2014; CADTH 2016; Cho 2010; Edmunds 2011; Francis 2015; Hall 2005; Hong 2013; Ito 2014; Lalakea 2003; Power 2015; Segal 2007; Suter 2009). All included observational studies as well as randomised trials. All reviews recognised a role for frenotomy when evidence indicates feeding difficulties or maternal nipple pain. However, review authors provided different interpretations of the strength of available evidence supporting frenotomy. Algar 2009, Bowley 2014, Cho 2010, Edmunds 2011, Hong 2013, Ito 2014, Lalakea 2003 and Segal 2007 concluded that evidence is sufficient to recommend frenotomy in an infant with breastfeeding problems and tongue-tie. CADTH 2016, Hall 2005, Power 2015, Suter 2009 and Francis 2015 concluded that frenotomy may be helpful and is safe but that definitive evidence is lacking. The most recent review (CADTH 2016) concluded that frenotomy is a safe procedure that leads to maternally perceived benefit in short-term breastfeeding outcomes, concurring with this review that objective improvement in symptoms and long-term benefit is less certain. CADTH 2016 concluded that it remains to be proved whether frenotomy provides meaningful improvement in breastfeeding difficulties, especially over the long term. Another recent review (Francis 2015) included the same five randomised trials included in this review, together with a retrospective cohort and 23 case series. Those review authors concluded that the strength of the evidence supporting frenotomy for improved breastfeeding outcomes is low to insufficient and acknowledged that maternal reported effect is greater. They identified gaps in the evidence including longer-term outcomes, applicability to infants born at non-tertiary centres, lack of consistency in diagnosis, effectiveness of non-surgical interventions and optimal age of intervention. Two reviews acknowledged that the optimal time of intervention is unknown (Bowley 2014; Power 2015). Bowley 2014 recognised that frenotomy can improve breastfeeding problems in a tongue-tied child but advised waiting at least two weeks before performing the procedure. All reviews concluded that the procedure appears to have a low complication rate when performed by trained operators. Many reviews reported that available studies have methodological flaws and risk of bias.

AUTHORS' CONCLUSIONS

Implications for practice

Frenotomy causes a short-term reduction in nipple pain among breastfeeding mothers and an inconsistent positive effect on infant breastfeeding. Owing to the small number of studies and the high incidence of methodological issues, definitive benefit has not been proven.

Implications for research

Additional high-quality randomised controlled trials are needed to confirm whether frenotomy in tongue-tied infants causes resolution of feeding difficulties with both short-term and longer-term follow-up. In such studies, frenotomy ideally should not be performed on control infants to allow long-term assessment of the effect of the intervention.

Other major uncertainties remain unaddressed.

- The effect of frenotomy on tongue-tied preterm infants has yet to be studied.
- The optimal age to perform frenotomy in infants remains unclear.
- The effect of tongue-tie on early infant weight gain and on maternal difficulties in establishing a breast milk supply remains to be clarified.
- It has yet to be demonstrated whether frenotomy in breastfeeding infants with tongue-tie and feeding difficulty leads to a longer duration of breastfeeding.
- Whether frenotomy is a painful procedure that requires analgesia or anaesthesia has yet to be established, as no study to date has quantified infant pain during and after frenotomy.

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REFERENCES

References to studies included in this review

Berry 2012 *{published data only}*
Berry J, Griffiths M, Westcott C. A double-blind, randomized, controlled trial of tongue-tie division and its immediate effect on breastfeeding. *Breastfeeding Medicine* 2012;**7**(3):189–93. PUBMED: 21999476]

Buryk 2011 *{published data only}*
Buryk M, Bloom D, Shope T. Efficacy of neonatal release of ankyloglossia. *Pediatrics* 2011;**128**(2):280–8. PUBMED: 21768318]

Dollberg 2006 *{published data only}*
Dollberg S, Botzer E, Grunis E, Mimouni FB. Immediate nipple pain relief after frenotomy in breast-fed infants with ankyloglossia: a randomized, prospective study. *Journal of Pediatric Surgery* 2006;**41**(9):1598–1600. PUBMED: 16952598]

Emond 2013 *{published data only}*
Emond A, Ingram J, Johnson D, Plair P, Whitelaw A, Copeland M, et al. Randomised controlled trial of early frenotomy in breastfed infants with mild-moderate tongue-tie. *Archives of Diseases in Childhood Fetal and Neonatal Edition* 2014;**99**(3):F189–95. PUBMED: 24249695]

Hogan 2005 *{published data only}*
Hogan M, Westcott C, Griffiths M. Randomized, controlled trial of division of tongue-tie in infants with feeding problems. *Journal of Paediatrics and Child Health* 2005;**41** (5-6):246–50. PUBMED: 15953322]

References to studies excluded from this review

Ngerncham 2013 *{published data only}*
Ngerncham S, Laohapensang M, Wongvisutdhi T, Ritjaroen Y, Painpichan N, et al. Lingual frenulum and effect on breastfeeding in Thai newborn infants. *Paediatrics and International Child Health* 2013;**33**(2):86–90. PUBMED: 23925281]

Yousefi 2015 *{published data only}*
Yousefi J, Namini FT, Raisolsadat SMA, Gillies R, Ashkezari A, Meara JG. Tongue-tie repair: z-plasty vs simple release. *Iran Journal of Otorhinolaryngology* 2015;**27**(79):127-35.

References to ongoing studies

Ricalde 2017 *{published data only}*
Ricalde P. Prospective Evaluation of Lingual Frenotomy in Newborns With Simultaneous Lip Tie for the Relief of Breastfeeding Pain. ClinicalTrials.gov.

Additional references

Algar 2009
Algar V. Question 2. Should an infant who is breastfeeding poorly and has a tongue tie undergo a tongue tie division? . *Archives of Disease in Childhood* 2009;**94**(11):911–2. [PUBMED: 19847004]

Amir 2006
Amir LH, James JP, Donath SM. Reliability of the Hazelbaker assessment tool for lingual frenulum function.

International Breastfeeding Journal 2006;**1**(1):3. [PUBMED: 16722609]

Bowley 2014
Bowley DM, Arul GS. Fifteen-minute consultation: the infant with a tongue tie. *Archives of Disease in Childhood. Education and Practice Edition* 2014;**99**(4):127-9. [PUBMED: 24419208]

CADTH 2016
Canadian Agency for Drugs and Technologies in Health. Frenectomy for the correction of ankyloglossia: a review of clinical effectiveness and guidelines. https://www.cadth.ca/frenectomy-correction-ankyloglossia-review-clinical-effectiveness-and-guidelines (accessed 15 July 2016): RC0785–000.

Cho 2010
Cho A, Kelsberg G, Safranek S. Clinical inquiries. When should you treat tongue-tie in a newborn?. *Journal of Family Practice* 2010;**59**(12):712a–b. [PUBMED: 21135930]

Coryllos 2004
Coryllos E, Genna CW, Salloum AC. Congenital tongue-tie and its impact on breastfeeding. AAP section on breastfeeding. http://www.aap.org/breastfeeding/files/pdf/BBM-8-27%20Newsletter.pdf 2004; Vol. Summer: (accessed 15 March 2013).

Edmunds 2011
Edmunds J, Miles SC, Fulbrook P. Tongue-tie and breastfeeding: a review of the literature. *Breastfeeding Review* 2011;**19**(1):19–26. [PUBMED: 21608523]

Francis 2015
Francis DO, Krishnaswami S, McPheeters M. Treatment of ankyloglossia and breastfeeding outcomes: a systematic review. *Pediatrics* 2015;**135**(6):e1458–66. [PUBMED: 25941303]

GRADEpro [Computer program]
McMaster University. GRADEpro [www.gradepro.org]. McMaster University, 2014.

Hall 2005
Hall DM, Renfrew MJ. Tongue tie. *Archives of Disease in Childhood* 2005;**90**(12):1211–5. [PUBMED: 16301545]

Hazelbaker 1993
Hazelbaker AK. The Assessment Tool for Lingual Frenulum Function (ATLFF): use in a lactation consultant private practise. Pasadena California, Pacific Oaks College, Thesis, 1993.

Higgins 2011
Higgins JPT, Green S (editors). *Cochrane Handbook for Systematic Reviews of Interventions Version 5.1.0*. The Cochrane Collaboration, updated March 2011.

Hong 2013
Hong P. Ankyloglossia (tongue-tie). *CMAJ* 2013;**185**(185): E128. [DOI: 10.1503/cmaj.120785

Ito 2014
Ito Y. Does frenotomy improve breast-feeding difficulties in infants with ankyloglossia?. *Pediatrics International* 2014;**56**(4):497-505. [PUBMED: 24978831]

Jackson 2012
Jackson R. Improving breastfeeding outcomes: the impact of tongue-tie. *Community Practitioner* 2012;**85**(6):42–4. [PUBMED: 22779397]

Jensen 1994
Jensen D, Wallace S, Kelsay P. LATCH: a breastfeeding charting system and documentation tool. *Journal of Obstetrics, Gynecologic, and Neonatal Nursing* 1994;**23**(1): 27–32. [PUBMED: 8176525]

Kotlow 2011
Kotlow L. Diagnosis and treatment of ankyloglossia and tied maxillary fraenum in infants using Er:YAG and 1064 diode lasers. *European Archives of Paediatric Dentistry* 2011; **12**(2):106–12. [PUBMED: 21473843]

Krechel 1995
Krechel SW, Bildner J. CRIES: a new neonatal postoperative pain measurement score-initial testing of validity and reliability. *Paediatric Anaesthesia* 1995;**5**(1):53–61. [PUBMED: 8521311]

Krol 2007
Krol DM, Keels MA. Oral conditions. *Pediatrics in Review* 2007;**28**(1):15–22. [PUBMED: 17197455]

Kumar 2012
Kumar M, Kalke E. Tongue-tie, breastfeeding difficulties and the role of frenotomy. *Acta Paediatrica* 2012;**101**(7): 687–9. [PUBMED: 22404175]

Lalakea 2002
Lalakea ML, Messner AH. Frenotomy and frenuloplasty: if, when and how. *Operative Techniques in Pediatric Otolaryngology* 2002;**13**(1):93–7. [DOI: 10.1053/otot.2002.32157

Lalakea 2003
Lalakea ML, Messner AH. Ankyloglossia: does it matter?. *Pediatric Clinics of North America* 2003;**50**:381-97.

Lawrence 1993
Lawrence J, Alcock D, McGrath P, Kay J, MacMurray SB, Dulberg C. The development of a tool to assess neonatal pain. *Neonatal Network* 1993;**12**(6):59–66. [PUBMED: 8413140]

Marchesan 2005
Marchesan IQ. Lingual frenulum: quantitative evaluation proposal. *International Journal of Orofacial Myology* 2005; **31**:39–48. [PUBMED: 16739711]

Mathews 1988
Mathews MK. Developing an instrument to assess infant breastfeeding behaviour in the early neonatal period. *Midwifery* 1988;**4**(4):154–65. [PUBMED: 3210979]

Melzack 1975
Melzack R. The short-form McGill Pain Questionnaire. *Pain* 1987;**30**(2):191–7. [PUBMED: 3670870]

Messner 2000a
Messner AH, Lalakea ML. Ankyloglossia: controversies in management. *International Journal of Pediatric Otorhinolaryngology* 2000;**54**(2-3):123–31. [PUBMED: 10967382]

Messner 2000b

Messner AH, Lalakea ML, Aby J, Macmahon J, Bair E. Ankyloglossia: incidence and associated feeding difficulties. *Archives of Otolaryngology-Head and Neck Surgery* 2000;**126**(1):36–9. [PUBMED: 10628708]

NICE 2005

National Institute of Health and Care Excellence (NICE). Division of ankyloglossia (tongue-tie) for breastfeeding. http://www.nice.org.uk/nicemedia/live/11180/31411/31411.pdf. (accessed 15 March 2013).

Obladen 2010

Obladen M. Much ado about nothing: two millennia of controversy on tongue-tie. *Neonatology* 2010;**97**(2):83–9. [PUBMED: 19707023]

Power 2015

Power RF, Murphy JF. Tongue-tie and frenotomy in infants with breastfeeding difficulties: achieving a balance. *Archives of Disease in Childhood* 2015;**100**(5):489-94. [PUBMED: 25381293]

RevMan 2014

The Cochrane Collaboration. Review Manager (RevMan). Version 5.3. The Cochrane Collaboration, 2014.

Ricke 2005

Ricke LA, Baker NJ, Madlon-Kay DJ, DeFor TA. Newborn tongue-tie: prevalence and effect on breast-feeding. *Journal of the American Board of Family Practice* 2005;**18**(1):1–7. [PUBMED: 15709057]

Schünemann 2013

Schünemann H, Broż ek J, Guyatt G, Oxman A, editors. GWG. GRADE Handbook for Grading Quality of Evidence and Strength of Recommendations. www.guidelinedevelopment.org/handbook. Updated October 2013.

Segal 2007

Segal LM, Stephenson R, Dawes M, Feldman P. Prevalence, diagnosis, and treatment of ankyloglossia. *Canadian Family Physician* 2007;**53**(6):1027–33. [PUBMED: 17872781]

Suter 2009

Suter VGA, Bornstein M. Ankyloglossia: facts and myths in diagnosis and treatment. *Journal of Periodontology* 2009;**80**(8):1204–19. [PUBMED: 19656020]

Taddio 1995

Taddio A, Nulman I, Koren BS, Stevens B, Koren G. A revised measure of acute pain in infants. *Journal of Pain and Symptom Management* 1995;**10**(6):456–63. [PUBMED: 7561228]

* *Indicates the major publication for the study*

CHARACTERISTICS OF STUDIES

Characteristics of included studies *[ordered by study ID]*

Berry 2012

Methods	Randomised blinded controlled trial performed between October 2003 and April 2004 at an English regional hospital
Participants	Infants younger than 4 months of age with symptoms of breastfeeding problems and tongue-tie
Interventions	Immediate frenotomy or non-division
Outcomes	Primary outcomes: subjective and objective improvement in feeding - feeding score (adapted from LATCH scoring system and Infant Breastfeeding Assessment Tool), maternal questioning, observer impression Secondary outcome: maternal pain scores
Notes	Non-division infants, then divided

Risk of bias

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	University of Southampton Medical Statistics and Computing Department provided computer-generated randomisation for 60 babies; an independent helper then placed the randomisation into sealed envelopes
Allocation concealment (selection bias)	Low risk	Sealed envelopes
Blinding of participants and personnel (performance bias) All outcomes	Low risk	Double-blinded
Blinding of outcome assessment (detection bias) All outcomes	Low risk	Double-blinded
Incomplete outcome data (attrition bias) All outcomes	Low risk	
Selective reporting (reporting bias)	High risk	Objective outcomes not reported numerically; said to be no different
Other bias	High risk	Non-divided babies offered division anyway; suggesting lack of equipoise

Buryk 2011

Methods	Single-blinded randomised controlled trial performed between December 2007 and December 2008 at an American regional military medical centre
Participants	Infants (< 30 days) with breastfeeding issues found to have significant tongue-tie (Hazelbaker Assessment Tool for Lingual Frenulum Function (ATLFF) scores: function scores > 11, appearance score < 8) Exclusion criteria described
Interventions	Frenotomy or sham procedure, immediately or within 1 to 2 weeks
Outcomes	Primary outcome: improvement in maternal nipple pain (McGill Pain scores) and ability to breastfeed (breastfeeding scores, Infant Breastfeeding Assessment Tool (IBFAT)) Secondary outcome: improvement in length of breastfeeding
Notes	Non-division infants were offered frenotomy 2 weeks later

Risk of bias

Bias	**Authors' judgement**	**Support for judgement**
Random sequence generation (selection bias)	Low risk	Computerised random number generator of blocks of 4 created by a statistician and implemented by a research assistant
Allocation concealment (selection bias)	Unclear risk	Did not state how investigators concealed allocation
Blinding of participants and personnel (performance bias) All outcomes	Low risk	Parents blinded but told the allocation after first feed post procedure. All medical professionals involved were not blinded
Blinding of outcome assessment (detection bias) All outcomes	Low risk	Longer-term follow-up less significant, as all but 1 control infant had a frenotomy
Incomplete outcome data (attrition bias) All outcomes	Low risk	
Selective reporting (reporting bias)	Low risk	
Other bias	High risk	All control infants were offered frenotomy

Dollberg 2006

Methods	Randomised cross-over study performed between December 2001 and September 2004 at an Israeli maternity hospital
Participants	Infants < 21 days old, all mothers had nipple pain; infants examined by neonatologist and found to have tongue-tie
Interventions	Cross-over study; randomised to sham, then feed, then frenotomy, then feed and vice versa
Outcomes	Pain and latch scores after frenotomy or sham
Notes	

Risk of bias

Bias	**Authors' judgement**	**Support for judgement**
Random sequence generation (selection bias)	Low risk	Computer-generated randomisation table
Allocation concealment (selection bias)	Low risk	Sealed opaque envelopes
Blinding of participants and personnel (performance bias) All outcomes	Low risk	Mothers and all treating personnel not blinded
Blinding of outcome assessment (detection bias) All outcomes	Low risk	Not blinded
Incomplete outcome data (attrition bias) All outcomes	Low risk	
Selective reporting (reporting bias)	High risk	Results in control group not reported in original manuscript have been since provided by study authors
Other bias	Low risk	

Emond 2013

Methods	Randomised feasibility trial of early frenotomy compared with usual care provided between December 2001 and September 2004 for infants with mild to moderate tongue-tie at a regional English maternity hospital
Participants	Mothers of babies with tongue-tie who were experiencing breastfeeding difficulties; Hazelbaker Assessment Tool for Lingual Frenulum Function (HTLFF) scores 6 to 12, latch score $\leq$ 8; infants with severe tongue-tie excluded and sent for frenotomy

Emond 2013 (*Continued*)

Interventions	Frenotomy or not Five days later, frenotomy offered to control group if symptoms persisted
Outcomes	Primary outcome: changes in latch scores at 5 days Secondary outcomes: changes in Infant Breastfeeding Assessment Tool (IBFAT) score and score on breastfeeding self-efficacy short form; changes in pain score on visual analogue scale
Notes	Four of the 99 frenotomies performed as initial procedure were repeated and did not sufficiently divide the frenulum

Risk of bias

Bias	**Authors' judgement**	**Support for judgement**
Random sequence generation (selection bias)	Low risk	Telephone-based block randomisation service stratified for sex and birth order All given routine breastfeeding support
Allocation concealment (selection bias)	Low risk	
Blinding of participants and personnel (performance bias) All outcomes	Low risk	Researchers were blinded, but mothers were not
Blinding of outcome assessment (detection bias) All outcomes	Low risk	
Incomplete outcome data (attrition bias) All outcomes	Low risk	
Selective reporting (reporting bias)	Low risk	
Other bias	High risk	First line of protocol states mothers of term infants with breastfeeding problems due to tongue-tie. Almost all infants had frenotomy at maternal request, suggesting lack of equipoise; almost all control infants were offered frenotomy, suggesting lack of equipoise

Hogan 2005

Methods	Randomised controlled trial performed between March and July 2002 that recruited participants from a regional English maternity hospital and 3 English birthing centres	
Participants	Infants 4 weeks of age	
Interventions	Frenotomy or intensive lactation support	
Outcomes	Subjective improvement in feeding	
Notes		
Risk of bias		
Bias	**Authors' judgement**	**Support for judgement**
Random sequence generation (selection bias)	Low risk	Random sequence was computer generated by our department of medical statistics and was placed in envelopes by a third party with no input from the 3 study authors
Allocation concealment (selection bias)	Low risk	Sealed envelope
Blinding of participants and personnel (performance bias) All outcomes	High risk	No blinding
Blinding of outcome assessment (detection bias) All outcomes	High risk	Study authors state that they did not have equipoise and provided no blinding
Incomplete outcome data (attrition bias) All outcomes	Low risk	
Selective reporting (reporting bias)	Low risk	
Other bias	High risk	All control infants were offered frenotomy

Characteristics of excluded studies *[ordered by study ID]*

Study	Reason for exclusion
Ngerncham 2013	Cross-sectional study
Yousefi 2015	Compared frenotomy versus frenuloplasty. Population included children up to 12 years of age

Characteristics of ongoing studies *[ordered by study ID]*

Ricalde 2017

Trial name or title	Prospective Evaluation of Lingual Frenotomy in Newborns With Simultaneous Lip Tie for the Relief of Breastfeeding Pain
Methods	Randomised controlled trial of newborns in maternal infant care areas at Tampa General Hospital
Participants	Term infants classified as having ankyloglossia via the HATLFF (Hazelbaker Assessment Tool for Lingual Frenulum Function) and a Class III or IV maxillary labial frenum
Interventions	Randomly assigned to 1 of 2 groups: Group A or Group B Group A will receive a sham procedure for intervention #1 and a lingual frenotomy procedure for intervention #2. Group B will receive a lingual frenotomy procedure for intervention #1 and a sham procedure for intervention #2 Newborns who continue to have difficulty with breastfeeding after both interventions will undergo intervention #3 - a labial frenotomy - and breastfeeding will be monitored afterwards
Outcomes	• Wong-Baker FACES Pain Rating Scale • LATCH score
Starting date	May 2015
Contact information	Pat Ricalde, MD, DDS Tampa General Hospital Tampa, Florida, United States, 33606 813-870-6000 ricalde@verizon.net Sponsor: University of South Florida
Notes	ClinicalTrials.gov identifier: NCT02141243

DATA AND ANALYSES

Comparison 1. Frenotomy versus no frenotomy or sham procedure

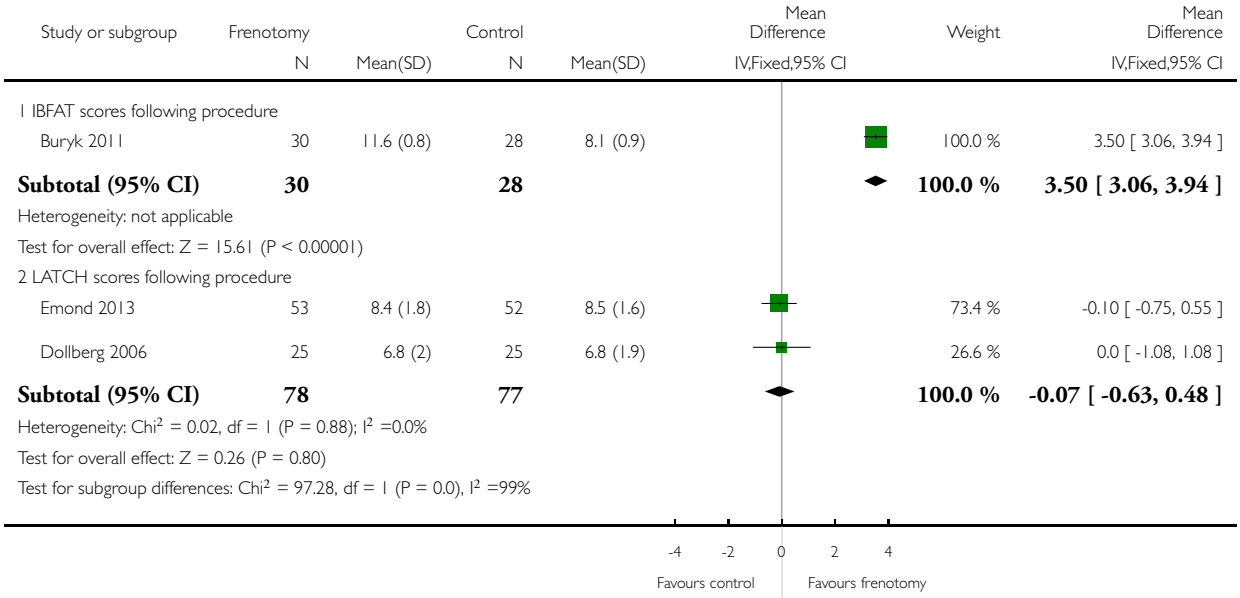
Outcome or subgroup title	No. of studies	No. of participants	Statistical method	Effect size
1 Infant breastfeeding assessed by a validated scale	3		Mean Difference (IV, Fixed, 95% CI)	Subtotals only
1.1 IBFAT scores following procedure	1	58	Mean Difference (IV, Fixed, 95% CI)	3.50 [3.06, 3.94]
1.2 LATCH scores following procedure	2	155	Mean Difference (IV, Fixed, 95% CI)	-0.07 [-0.63, 0.48]
2 Infant breastfeeding assessed by a validated scale 2 to 7 days following procedure	1	105	Mean Difference (IV, Fixed, 95% CI)	-0.10 [-0.75, 0.55]
3 Maternal nipple pain assessed by a validated pain scale	4		Mean Difference (IV, Fixed, 95% CI)	Subtotals only
3.1 Visual analogue pain scale	3	183	Mean Difference (IV, Fixed, 95% CI)	-0.74 [-1.35, -0.13]
3.2 SF-MPQ pain scale	1	58	Mean Difference (IV, Fixed, 95% CI)	-8.6 [-9.37, -7.83]
4 Qualitative assessment of infant feeding by parental survey performed within 48 hours of procedure	2	114	Risk Ratio (M-H, Fixed, 95% CI)	3.48 [2.18, 5.56]
5 Excessive bleeding at the time or within 24 hours of frenotomy (as determined by study investigators)	5	302	Risk Difference (M-H, Fixed, 95% CI)	0.0 [-0.03, 0.03]
6 Infection at the site of frenotomy requiring treatment with antibiotics within 7 days of procedure	5	302	Risk Ratio (M-H, Fixed, 95% CI)	0.0 [0.0, 0.0]
7 Damage to the tongue and/or submandibular ducts noted within 7 days of procedure (as determined by study investigators)	5	302	Risk Ratio (M-H, Fixed, 95% CI)	0.0 [0.0, 0.0]

Analysis 1.1. Comparison 1 Frenotomy versus no frenotomy or sham procedure, Outcome 1 Infant breastfeeding assessed by a validated scale.

Review: Frenotomy for tongue-tie in newborn infants

Comparison: 1 Frenotomy versus no frenotomy or sham procedure

Outcome: 1 Infant breastfeeding assessed by a validated scale



Analysis 1.2. Comparison 1 Frenotomy versus no frenotomy or sham procedure, Outcome 2 Infant breastfeeding assessed by a validated scale 2 to 7 days following procedure.

Review: Frenotomy for tongue-tie in newborn infants

Comparison: 1 Frenotomy versus no frenotomy or sham procedure

Outcome: 2 Infant breastfeeding assessed by a validated scale 2 to 7 days following procedure

Study or subgroup	Frenotomy N	Mean(SD)	Control N	Mean(SD)	Mean Difference IV,Fixed,95% CI	Weight	Mean Difference IV,Fixed,95% CI
Emond 2013	53	8.4 (1.8)	52	8.5 (1.6)		100.0 %	-0.10 [-0.75, 0.55]
Total (95% CI)	**53**		**52**			**100.0 %**	**-0.10 [-0.75, 0.55]**

Heterogeneity: not applicable

Test for overall effect: Z = 0.30 (P = 0.76)

Test for subgroup differences: Not applicable

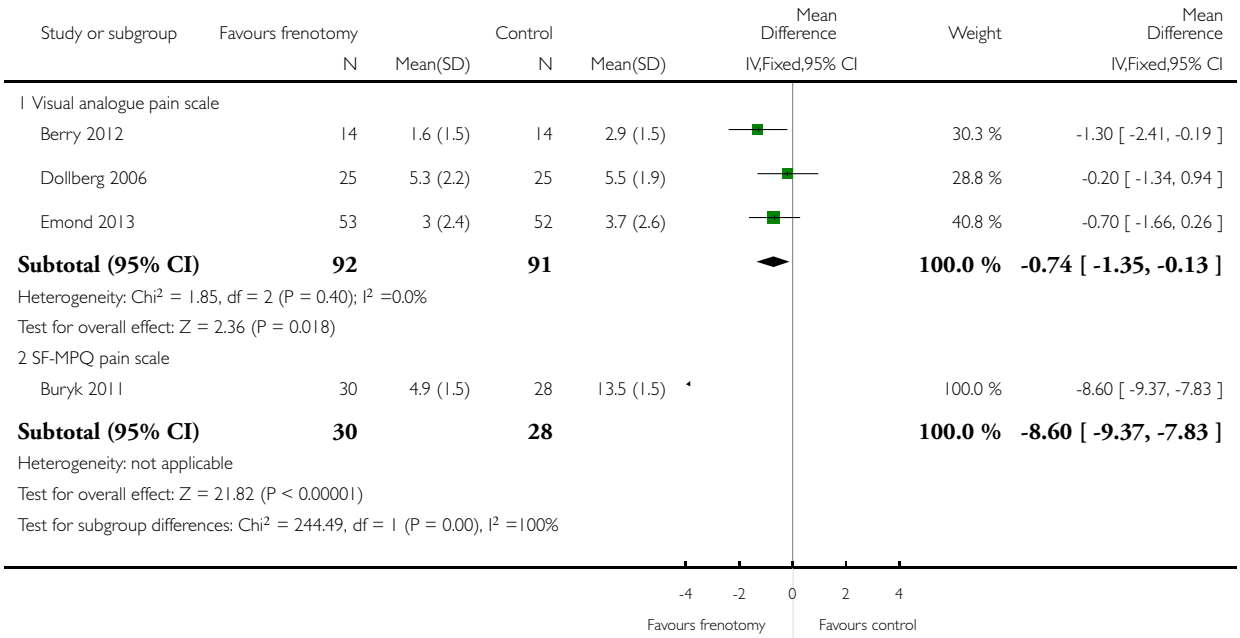
Analysis 1.3. Comparison 1 Frenotomy versus no frenotomy or sham procedure, Outcome 3 Maternal nipple pain assessed by a validated pain scale.

Review: Frenotomy for tongue-tie in newborn infants

Comparison: 1 Frenotomy versus no frenotomy or sham procedure

Outcome: 3 Maternal nipple pain assessed by a validated pain scale

Study or subgroup	Favours frenotomy		Control		Mean Difference	Weight	Mean Difference
	N	Mean(SD)	N	Mean(SD)	IV,Fixed,95% CI		IV,Fixed,95% CI
1 Visual analogue pain scale							
Berry 2012	14	1.6 (1.5)	14	2.9 (1.5)		30.3 %	-1.30 [-2.41, -0.19]
Dollberg 2006	25	5.3 (2.2)	25	5.5 (1.9)		28.8 %	-0.20 [-1.34, 0.94]
Emond 2013	53	3 (2.4)	52	3.7 (2.6)		40.8 %	-0.70 [-1.66, 0.26]
Subtotal (95% CI)	**92**		**91**			**100.0 %**	**-0.74 [-1.35, -0.13]**
Heterogeneity: Chi^2 = 1.85, df = 2 (P = 0.40); I^2 =0.0%							
Test for overall effect: Z = 2.36 (P = 0.018)							
2 SF-MPQ pain scale							
Buryk 2011	30	4.9 (1.5)	28	13.5 (1.5)		100.0 %	-8.60 [-9.37, -7.83]
Subtotal (95% CI)	**30**		**28**			**100.0 %**	**-8.60 [-9.37, -7.83]**
Heterogeneity: not applicable							
Test for overall effect: Z = 21.82 (P < 0.00001)							
Test for subgroup differences: Chi^2 = 244.49, df = 1 (P = 0.00), I^2 =100%							



Analysis 1.4. Comparison 1 Frenotomy versus no frenotomy or sham procedure, Outcome 4 Qualitative assessment of infant feeding by parental survey performed within 48 hours of procedure.

Review: Frenotomy for tongue-tie in newborn infants

Comparison: 1 Frenotomy versus no frenotomy or sham procedure

Outcome: 4 Qualitative assessment of infant feeding by parental survey performed within 48 hours of procedure

Study or subgroup	Frenotomy n/N	Control n/N	Risk Ratio M-H,Fixed,95% CI	Weight	Risk Ratio M-H,Fixed,95% CI
Berry 2012	21/27	14/30		93.1 %	1.67 [1.08, 2.57]
Hogan 2005	27/28	1/29		6.9 %	27.96 [4.07, 192.12]
Total (95% CI)	**55**	**59**		**100.0 %**	**3.48 [2.18, 5.56]**

Total events: 48 (Frenotomy), 15 (Control)

Heterogeneity: Chi^2 = 15.63, df = 1 (P = 0.00008); I^2 =94%

Test for overall effect: Z = 5.22 (P < 0.00001)

Test for subgroup differences: Not applicable

Analysis 1.5. Comparison 1 Frenotomy versus no frenotomy or sham procedure, Outcome 5 Excessive bleeding at the time or within 24 hours of frenotomy (as determined by study investigators).

Review: Frenotomy for tongue-tie in newborn infants

Comparison: 1 Frenotomy versus no frenotomy or sham procedure

Outcome: 5 Excessive bleeding at the time or within 24 hours of frenotomy (as determined by study investigators)

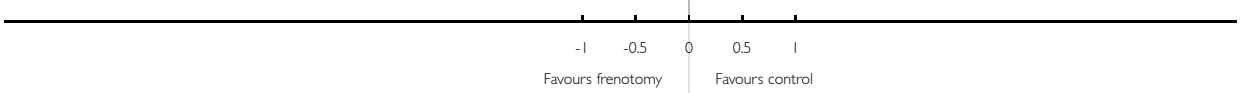
Study or subgroup	Experimental n/N	Control n/N	Risk Difference M-H,Fixed,95% CI	Weight	Risk Difference M-H,Fixed,95% CI
Emond 2013	0/53	0/52		34.8 %	0.0 [-0.04, 0.04]
Hogan 2005	0/28	0/29		18.9 %	0.0 [-0.07, 0.07]
Buryk 2011	0/30	0/28		19.2 %	0.0 [-0.06, 0.06]
Berry 2012	0/27	0/30		18.9 %	0.0 [-0.07, 0.07]
Dollberg 2006	0/14	0/11		8.2 %	0.0 [-0.15, 0.15]
Total (95% CI)	**152**	**150**		**100.0 %**	**0.0 [-0.03, 0.03]**

Total events: 0 (Experimental), 0 (Control)

Heterogeneity: $Chi^2 = 0.0$, $df = 4$ ($P = 1.00$); $I^2 = 0.0\%$

Test for overall effect: $Z = 0.0$ ($P = 1.0$)

Test for subgroup differences: Not applicable



Analysis 1.6. Comparison 1 Frenotomy versus no frenotomy or sham procedure, Outcome 6 Infection at the site of frenotomy requiring treatment with antibiotics within 7 days of procedure.

Review: Frenotomy for tongue-tie in newborn infants

Comparison: 1 Frenotomy versus no frenotomy or sham procedure

Outcome: 6 Infection at the site of frenotomy requiring treatment with antibiotics within 7 days of procedure

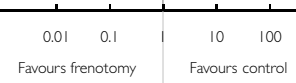
Study or subgroup	Frenotomy n/N	Control n/N	Risk Ratio M-H,Fixed,95% CI	Weight	Risk Ratio M-H,Fixed,95% CI
Berry 2012	0/28	0/29			Not estimable
Buryk 2011	0/53	0/52			Not estimable
Dollberg 2006	0/14	0/11			Not estimable
Emond 2013	0/30	0/28			Not estimable
Hogan 2005	0/27	0/30			Not estimable
Total (95% CI)	**152**	**150**			**Not estimable**

Total events: 0 (Frenotomy), 0 (Control)

Heterogeneity: not applicable

Test for overall effect: not applicable

Test for subgroup differences: Not applicable



Analysis 1.7. Comparison 1 Frenotomy versus no frenotomy or sham procedure, Outcome 7 Damage to the tongue and/or submandibular ducts noted within 7 days of procedure (as determined by study investigators).

Review: Frenotomy for tongue-tie in newborn infants

Comparison: 1 Frenotomy versus no frenotomy or sham procedure

Outcome: 7 Damage to the tongue and/or submandibular ducts noted within 7 days of procedure (as determined by study investigators)

Study or subgroup	Experimental n/N	Control n/N	Risk Ratio M-H,Fixed,95% CI	Weight	Risk Ratio M-H,Fixed,95% CI
Berry 2012	0/28	0/29			Not estimable
Buryk 2011	0/53	0/52			Not estimable
Dollberg 2006	0/14	0/11			Not estimable
Emond 2013	0/30	0/28			Not estimable
Hogan 2005	0/27	0/30			Not estimable
Total (95% CI)	**152**	**150**			**Not estimable**

Total events: 0 (Experimental), 0 (Control)

Heterogeneity: not applicable

Test for overall effect: not applicable

Test for subgroup differences: Not applicable

HISTORY

Date	Event	Description
10 July 2008	Amended	Converted to new review format

CONTRIBUTIONS OF AUTHORS

CO'D and DB wrote the first draft of the protocol.

JO'S and JF wrote subsequent drafts of the protocol.

PD, SJ, DT and CO'D commented on and reviewed the protocol.

JO'S and JF wrote the review. PD, SJ, DT and CO'D commented on and reviewed the review.

DECLARATIONS OF INTEREST

None declared.

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DIFFERENCES BETWEEN PROTOCOL AND REVIEW

We added to this review the methods and plan for 'Summary of findings' tables and GRADE recommendations, which were not included in the original protocol.

INDEX TERMS

Medical Subject Headings (MeSH)

*Breast Feeding [adverse effects]; Ankyloglossia [*surgery]; Gestational Age; Lingual Frenum [*surgery]; Mastodynia [etiology]; Nipples; Pain Measurement; Randomized Controlled Trials as Topic

MeSH check words

Female; Humans; Infant, Newborn