Specific Care Question:

Is frenulectomy beneficial in infants with ankyloglossia?

Question Originator:

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Plain Language Summary from The Office of Evidence Based Practice:

Ankyloglossia (tongue-tie) with or without lip-tie is a congenital oral deformity. The lingual frenulum (tissue connecting tongue to floor and/or roof of mouth) is abnormally short, thick, tight, or has developed anterior instead of posterior. Stated effects of ankyloglossia in breastfeeding infants include poor latch and maternal nipple pain.

Based on very low quality evidence, a weak recommendation is made to consider tongue-tie division, specifically if infant feeding difficulties and/or maternal nipple pain are present. Very low quality studies report improvement in these two factors after frenulectomy (Power & Murphy, 2015). From the systematic review by Francis et al.,(2015) tongue-tie division may be considered when ankyloglossia interferes with feeding or is thought to be a factor in maternal nipple pain. Data is not available for infants with craniofacial abnormalities, Pierre Robin syndrome or sequence, Down syndrome, or prematurity (<37 weeks PMA). Data from two studies included in Francis et al. (2015), are included in the in Figures 1-4. As you can see, the number of subjects recruited is very low with 56 subjects in Berry, Griffiths, & Westcott (2012) and 58 subjects in Buryk, Bloom, & Shope (2011). Maternal perception of feeding improvement was significantly greater when infants had the tongue-tie divided (See Figure 1) but independent observers found no difference (See Figure 2). Buryk, Bloom, & Shope (2011) reported no difference in SF-MPQ pain score or IBFAT feeding score, which were both subjective reports by mothers (See Figures 3 and 4). Harms related to the procedure have been reported as low and are bleeding and pain. The existing literature does not clearly assess long-term success of breast feeding duration after frenulectomy because parents often decide to have their child's frenulectomy divided after the study(ies) is completed..

Literature Review

The literature search identified three papers that are included in this synthesis. Infant age ranged from 6 days to 4 months old. Most studies found subjective outcome measures support tongue-tie division; e.g., maternal perception of breastfeeding efficacy and reports of nipple pain resolution. However, objective outcome measures such as an observer's analysis of latch success rarely demonstrated statistically significant results. In most studies, mothers in groups of infants who were not selected for tongue-tie division were given the option to operate after the conclusion of the study period. Most of these mothers opted for intervention, which eliminated the possibility of long-term comparisons of breast feeding outcomes like time to weaning. In the one study examining non-breastfeeding outcomes such as speech and the ability to clean the mouth, the data failed to support tongue-tie



division (Chinnadurai et al., 2015).

There is a research gap in evaluating harms for tongue-tie division. The frenulum is not significantly vascular, so bleeding has been reported to be mild. The tissue also has few nerve endings, so infants' subsequent crying after division is resolved by immediate breastfeeding (Francis et al., 2015). Procedural pain has not been evaluated.

EBP Scholar's responsible for analyzing the literature:

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EBP team member responsible for reviewing, synthesizing, and developing this literature:

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Search Strategy and Results:

PubMed: "Lingual Frenum/surgery" [Mesh] OR (("frenectomy" OR "frenulectomy") AND ("lingual" OR "ankyloglossia" OR "tongue")) Filters: 5 years, Infant: birth-23 months Results: 39 articles. Five articles were selected. One paper is a systematic review ((Francis et al., 2015)) and includes (Berry et al., 2012) and (Buryk et al., 2011). Data from the former studies are included in the Figures.

Studies included in this review:

Chinnadurai et al., (2015)

Francis et al., (2015)

Power & Murphy, (2015)

Studies not included in this review with rationale for exclusion:

Study	Rationale for exclusion
Berry et al., (2012)	Included in Francis 2015, but data from the study is included in Figures 1 and 2
Buryk et al., (2011)	Included in Francis 2015, but data from the study is included in Figures 3 and 4
Emond et al., (2014)	Included in Francis 2015

Method Used for Appraisal and Synthesis:

The Cochrane Collaborative computer program, Review Manager (RevMan 5.3.5), was used to synthesize the three included studies. The details of the three excluded studies are included for review.

Final: January 28 2016



Characteristics of included studies:

Tables:

Chinnadurai 2015

Methods	Systematic review: 2 randomized controlled trials, 2 cohort studies, 11 case series								
Participants	Setting: Variety Inclusion criteria: Children 0-18 years with tongue tie or tongue-tie and lip-tie Exclusion criteria: Van der Woulde syndrome, Pierre Robin syndrome, Down syndrome, craniofacial abnormalities, premature infants								
Interventions	Surgical interventions: simple anterior frenotomy, laser frenulectomy, posterior frenulectomy, Z-plasty repair Nonsurgical interventions:complementary and alternative medicine therapies (craniosacral therapy, myofascial release, chiropractic therapies), lactation intervention, speech therapy, physical therapy, oral motor therapy, and stretching exercises/therapy								
Outcomes	Speech: assessments of articulation and intelligibility Feeding: maternal report of bottle-feeding success, ability to use tongue to eat ice cream and clean the mouth Social: ability to clean teeth with tongue, licking outside of lips, eating ice cream								
Notes	Speech & articulation outcomes - strength of evidence insufficient to support surgical interventions Bottle feeding/use of tongue for licking and hygiene outcomes - data is insufficient to support surgical interventions Social outcomes - strength of evidence insufficient to support surgical interventions Harms - generally mild to nonexistent, included bleeding that resolved quickly. No harms of treatment associated with older children's care reported.								

Francis 2015

Methods	Systematic review - 6 randomized controlled trials (poor to good quality), 3 cohort studies (all poor quality), 33
	case series, 15 case reports, 1 unpublished thesis



Participants	Setting: Inpatient or outpatient pediatric care, operating room, newborn nursery or NICU, ENT clinic, primary care outpatient, dental office, breastfeeding medicine clinic. Studies included American, Canadian, European and Asian settings. Inclusion criteria: Children ages 0–18 with tongue-tie or tongue-tie with concomitant lip-tie. English studies. Exclusion criteria: Van der Woude syndrome, Pierre Robin syndrome or sequence, Down syndrome, or craniofacial abnormalities, and premature babies (<37 weeks of gestation). Non-English studies.
Interventions	Intervention groups: Surgical interventions (simple anterior frenotomy, frenulotomy, or frenectomy; laser frenotomy, or frenulectomy; posterior frenulectomy; Z-plasty repair) Nonsurgical treatments, including complementary and alternative medicine therapies (e.g., craniosacral therapy, myofascial release, and other chiropractic therapies), lactation intervention, speech therapy, physical therapy, oral motor therapy, and stretching exercises/therapy Control groups: Other surgical approach, non-surgical interventions including lactation intervention, speech therapy; physical/occupational therapy oral motor therapy, and stretching exercises/therapy; observation; complementary and alternative medicine (CAM) therapies (e.g. craniosacral therapy); placebo (sham therapy)
Outcomes	 For neonates and infants: Intermediate outcomes: Maternal nipple pain, ability to latch and maintain latch, tongue mobility, alleviation of aerophagia Final outcomes: Duration of breastfeeding, failure to thrive, infant weight gain, oral and oropharyngeal dysphagia For children through 18 years of age: Intermediate outcomes: tongue mobility, feeding sequelae Final outcomes: articulation disorder, feeding sequelae, oral hygiene, oral and oropharyngeal dysphagia, orthodontic problems, psychological outcomes, social concerns (e.g., kissing)
Notes	Breastfeeding outcomes - small body of evidence suggests that frenotomy may be associated with improvements in breastfeeding as reported by mothers, and potentially in nipple pain, but strength of evidence is generally low to insufficient. Feeding outcomes - strength of evidence insufficient to support surgical intervention



	Speech outcomes - strength of evidence insufficient to support surgical intervention
	Social concerns related to tongue mobility - strength of evidence insufficient to support surgical intervention
	Harms - moderate strength of evidence for minimal and short-lived bleeding. Insufficient strength of evidence
	for pain and reoperation.

Power 2014

Methods	Systematic review - 4 systematic reviews and 5 randomized controlled trials
Participants	Setting: Inpatient or outpatient pediatric care, operating room, newborn nursery or NICU, ENT clinic, primary care outpatient, dental office, breastfeeding medicine clinic. Studies included American, Canadian, European and Asian settings. Inclusion criteria: Infants less than 3 months old with tongue-tie and/or feeding problems. English studies Exclusion criteria: Infants with cleft lip, cleft palate, oral anomalies, and neuromuscular disorders. Non-English studies
Interventions	Intervention groups: frenotomy Control groups: placebo/sham/lactation support
Outcomes	Efficacy of breastfeeding after frenotomy; various outcome measures
Notes	Results: Evidence suggests a subjective improvement in breastfeeding after frenotomy.

Figures:



Figure 1. Division vs. Placebo, Outcome: Maternal Perception of Improved Feeding

Division		ivision Placebo			_	Odds Ratio	Odds Ratio
Study or Subgroup	Events	Total	Events	Total	Weight	M-H, Fixed, 95% CI	CI M-H, Fixed, 95% CI
Berry 2012	21	27	14	30	100.0%	4.00 [1.26, 12.72]	
Total (95% CI)		27		30	100.0%	4.00 [1.26, 12.72]	
Total events	21		14				
Heterogeneity: Not ap Test for overall effect:		P = 0.0	2)				0.01 0.1 1 10 100 Favors placebo Favors division

Figure 2. Division vs. Placebo, Outcome: Observer Perception of Improved Feeding

	Divisi	on	Placel	bo		Odds Ratio	Odds Ratio
Study or Subgroup	Events	Total	Events	Total	Weight	M-H, Fixed, 95% CI	M-H, Fixed, 95% CI
Berry 2012	13	26	12	30	100.0%	1.50 [0.52, 4.33]	-
Total (95% CI)		26		30	100.0%	1.50 [0.52, 4.33]	
Total events	13		12				
Heterogeneity: Not appropriate the control of the c		P = 0.4	5)				0.01 0.1 1 10 100 Favors placebo Favors division



Figure 3. Division vs. Placebo, Outcome: SF-MPQ, Pain Score

	Division			Placebo			Mean Difference			Mean Difference			
Study or Subgroup	Mean	SD	Total	Mean	SD	Total	Weight	IV, Fixed, 95% CI		IV, F	xed, 95	% CI	
Buryk 2011	16.8	10.6	30	19.2	9.9	28	100.0%	-2.40 [-7.68, 2.88]					
Total (95% CI)			30			28	100.0%	-2.40 [-7.68, 2.88]					
Heterogeneity: Not ap Test for overall effect:	•	(P = 0).37)						-20	-10 Favors interventi	0 on Fav	10 /ors placebo	20

Figure 4. Division vs. Placebo, Outcome: IBFAT, Feeding Score

Division		Pla	Placebo			Mean Difference	Mean Difference		
Study or Subgroup	Mean	SD	Total	Mean	SD	Total	Weight	IV, Fixed, 95% CI	IV, Fixed, 95% CI
Buryk 2011	9.3	3.7	30	8.5	3.8	28	100.0%	0.80 [-1.13, 2.73]	-
Emond 2013	0	0	0	0	0	0		Not estimable	
Total (95% CI)			30			28	100.0%	0.80 [-1.13, 2.73]	
Heterogeneity: Not ap	plicable								-4 -2 0 2 4
Test for overall effect:	Z = 0.81	(P =	0.42)						-4 -2 0 2 4 Favors placebo Favors division



References

Included:

- Chinnadurai, S., Francis, D. O., Epstein, R. A., Morad, A., Kohanim, S., & McPheeters, M. (2015). Treatment of ankyloglossia for reasons other than breastfeeding: a systematic review. *Pediatrics*, *135*(6), e1467-1474. doi:10.1542/peds.2015-0660
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- Power, R. F., & Murphy, J. F. (2015). Tongue-tie and frenotomy in infants with breastfeeding difficulties: achieving a balance. *Arch Dis Child*, 100(5), 489-494. doi:10.1136/archdischild-2014-306211

Excluded:

- Berry, J., Griffiths, M., & Westcott, C. (2012). A double-blind, randomized, controlled trial of tongue-tie division and its immediate effect on breastfeeding. *Breastfeed Med*, 7(3), 189-193. doi:10.1089/bfm.2011.0030
- Buryk, M., Bloom, D., & Shope, T. (2011). Efficacy of neonatal release of ankyloglossia: a randomized trial. *Pediatrics*, 128(2), 280-288. doi:10.1542/peds.2011-0077
- Emond, A., Ingram, J., Johnson, D., Blair, P., Whitelaw, A., Copeland, M., & Sutcliffe, A. (2014). Randomised controlled trial of early frenotomy in breastfed infants with mild-moderate tongue-tie. *Arch Dis Child Fetal Neonatal Ed*, 99(3), F189-195. doi:10.1136/archdischild-2013-305031

