

FINAL Key Questions and Background

Frenotomy and Frenectomy with Breastfeeding Support

Background

Estimates of ankyloglossia (i.e., tongue-tie) vary from <1% to about 11%, with prevalence more common among males than females.¹⁻³ Reasons for the wide variance in prevalence arise from unclear diagnostic methods, which may include visual inspection of the oral anatomy, assessment of functional impairment and decreased mobility, and the effect on mothers during breastfeeding (such as nipple pain).³ Ankyloglossia may be most commonly anterior, that is, where the frenum attaches near the tip of the tongue and is visible, or less commonly posterior, where the frenulum is attached further back on the tongue and may be harder to see.⁴ Of note, there is no consensus as to the definition of “posterior ankyloglossia” including whether this represents a distinct clinical entity.^{5, 6} Categories of severity have been proposed that rely on free tongue length⁷ and additional anatomical features (thickness, notching),⁸ but the relationship between these categories and breastfeeding difficulty have not been established.⁹ As a result, additional functional assessments of breastfeeding such as the LATCH index, Infant Breastfeeding Assessment Tool (IBFAT), or Frenotomy Decision Rule for Breastfeeding Infants (FDRBI) may be needed.⁹ The absence of validated diagnostic criteria creates uncertainty around the threshold for management.³

Outcomes potentially associated with untreated ankyloglossia include breastfeeding difficulties that may result in restricted weight gain in the infant,¹⁰⁻¹³ speech difficulties and problems with dentition,^{14, 15} and maternal pain, reduced milk supply, or incomplete emptying in the mother that may result in infections.^{16, 17}

Diagnosis of ankyloglossia and rates of frenotomy have increased sharply over the past 2 decades. Diagnoses of ankyloglossia in the US increased from 3,377 in 2004 to 13,200 in 2019 and lingual frenotomy to address lip-tie increased from 1,483 in 2004 to 6,213 in 2019.¹⁸

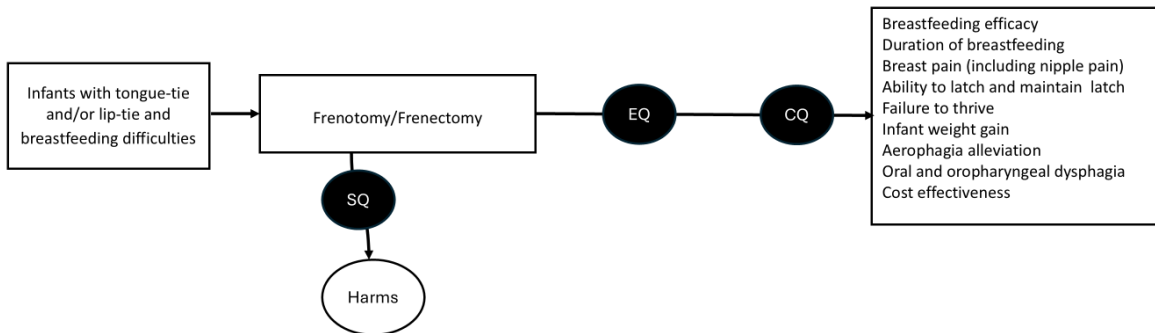
Policy Context

The State of Washington Health Care Authority selected frenectomy and frenotomy for breastfeeding support for a health technology assessment (HTA) because of high concerns for efficacy, and medium concerns for safety and cost.

Scope of this HTA

The analytic framework (**Figure 1**), research questions, and key study selection criteria (**Table 1**) are listed in this section.

Figure 1. Analytic Framework Depicting Scope of this Health Technology Assessment



Abbreviations: CQ = cost question; EQ = efficacy question; SQ = safety question.

Research Questions

Efficacy Question. What is the effectiveness and comparative effectiveness of frenotomy or frenectomy for tongue-tie and/or lip-tie on breastfeeding outcomes?

Safety Question. What are the harms of frenotomy or frenectomy for tongue-tie and/or lip-tie as a support for breastfeeding?

Cost Question. What is the cost-effectiveness of frenotomy or frenectomy for tongue-tie and/or lip-tie for breastfeeding support?

Study Selection Criteria

Table 1 provides the study selection criteria we will use to include studies in the HTA.

Table 1. Proposed Population, Intervention, Comparator, Outcome, Timing, and Setting for Health Technology Assessment on Frenotomy or Frenectomy with Breastfeeding Support

Domain	Included	Excluded
Population	Breastfeeding newborns with tongue-tie and/or lip-tie	<ul style="list-style-type: none"> • Infants with physical/anatomic comorbidities, such as hypotonia • Infants with Pierre Robin syndrome or sequence, Down syndrome, or craniofacial or airway abnormalities (i.e., cleft palate) • Infants born at less than <37 weeks of gestation
Intervention	Frenotomy, frenectomy, frenulotomy, frenulopasty, or z-plasty to improve breastfeeding using all methods (i.e., scissors, lasers)	Frenotomy, frenectomy, frenulotomy, or z-plasty done for indications other than breastfeeding support
Comparator	<ul style="list-style-type: none"> • EQ: All comparators including other surgical approaches, sham surgery, non-surgical interventions (i.e., lactation intervention, speech therapy, physical/occupational therapy, oral motor 	<ul style="list-style-type: none"> • EQ: No comparator group • SQ: N/A • CQ: No comparator group

Domain	Included	Excluded
	<p>therapy, and stretching exercises/therapy), complementary and alternative medicine (CAM) therapies (e.g. craniosacral therapy), observation only</p> <ul style="list-style-type: none"> • SQ: No comparator necessary • CQ: Any comparator 	
Outcomes	<ul style="list-style-type: none"> • *EQ: Breastfeeding, including latch, nipple pain, nipple excoriations, nipple infections (mastitis), weight gain, aerophagia, swallowing function, failure to thrive, milk transfer, low milk supply, breastfeeding cessation/duration of breastfeeding, and other feeding issues • SQ: Any harms, including excessive bleeding, airway obstruction, pain, transient poor feeding secondary to discomfort, dysphagia, complications related to dysphagia such as aspiration pneumonia, surgical site infection, nerve damage, salivary gland damage, ranulae, scarring, soft tissue damage, oral aversion, readherence of tongue- or lip-tie, need for further surgery/revision, ED visits, hospitalizations, extension of current hospitalization. • CQ: cost effectiveness or cost-utility 	<ul style="list-style-type: none"> • Outcomes not listed as eligible • Cost-effectiveness based on cost inputs from countries other than the U.S.
Timing	<ul style="list-style-type: none"> • EQ: Outcomes measured after intervention/comparator through 12 months of age • SQ: no time limitation • CQ: no time limitations 	Outcomes measured after 12 months of age
Setting	Inpatient or outpatient pediatric care, operating room, newborn nursery or NICU, ENT clinic, primary care outpatient, dental office, or breastfeeding medicine clinics in countries categorized as “very high” on the 2023/2024 UN Human Development Index. ¹⁹	Studies conducted in countries not categorized as “very high” on the 2023/2024 UN Human Development index. ¹⁹
Study Design	<ul style="list-style-type: none"> • EQ: RCTs, nonrandomized controlled trials, prospective and retrospective cohort studies, cross over studies, and case control studies • SQ: Same as for EQ plus case series • CQ: cost-effectiveness or cost-utility studies 	<ul style="list-style-type: none"> • EQ: Case reports, case-series, SRs, and qualitative studies • SQ: SRs, qualitative studies, and all study designs not already specified • CQ: Studies that use non-U.S. based cost inputs. • EQ, SQ, and CQ: Relevant SRs will be excluded but will be hand searched to identify potentially eligible primary studies
Language	<ul style="list-style-type: none"> • English 	<ul style="list-style-type: none"> • Non-English
Publication Type	<ul style="list-style-type: none"> • Original research 	<ul style="list-style-type: none"> • Editorial, commentaries, narrative reviews, or letters

*Dependent on the volume of captured EQ evidence, evidence synthesis and grading may be limited to validated measures.

Abbreviations: CQ =cost question; ENT = ear, nose and throat; EQ = efficacy question; N/A = not applicable; NICU = neonatal intensive care unit; RCT = randomized control trial; SQ = safety question; SR = systematic review.

Notes: ^a Countries identified as *very high* on the 2023/2024 UN Human Development Index: Andorra, Antigua and Barbuda, Argentina, Australia, Austria, Bahamas, Bahrain, Barbados, Belarus, Belgium, Brunei Darussalam, Canada, Chile, Costa Rica, Croatia, Cyprus, Czechia, Denmark, Estonia, Finland, France, Georgia, Germany, Greece, Hong Kong, China (SAR), Hungary, Iceland, Ireland, Israel, Italy, Japan, Kazakhstan, Korea (Republic of), Kuwait, Latvia, Liechtenstein, Lithuania, Luxembourg, Malaysia, Malta, Montenegro, Netherlands, New Zealand, Norway, Oman, Panama, Poland, Portugal, Qatar, Romania, Russian Federation, Saint Kitts and Nevis, San Marino, Saudi Arabia, Serbia, Seychelles, Singapore, Slovakia, Slovenia, Spain, Sweden, Switzerland, Thailand, Trinidad and Tobago, Türkiye, United Arab Emirates, United Kingdom, United States, Uruguay.¹⁹

What is Excluded from this HTA

This review will not include studies published in languages other than English or studies conducted in countries less than ‘very high’ on the 2023/2024 United Nations Human Development Index.¹⁹ This review will not include studies that examine frenotomies and frenectomies performed for reasons other than breastfeeding support (e.g., articulation). This review will also not include studies conducted among infants with major comorbidities, other abnormalities, or who were born at less than 37 weeks gestation. This review will exclude studies with no comparison group for the EQ.

References

1. Hazelbaker AK, Baeza C, Genna CW, et al. Incidence and Prevalence of Tongue-Tie. *Clin Lactation*. 2017(3):89-92. doi: 10.1891/2158-0782.8.3.89.
2. Sedano HO. Congenital oral anomalies in argentinian children. *Community Dent Oral Epidemiol*. 1975 Mar;3(2):61-3. doi: 10.1111/j.1600-0528.1975.tb00281.x. PMID: 1056288.
3. Segal LM, Stephenson R, Dawes M, Feldman P. Prevalence, diagnosis, and treatment of ankyloglossia: methodologic review. *Can Fam Physician*. 2007 Jun;53(6):1027-33. PMID: 17872781.
4. Hong P, Lago D, Seargeant J, et al. Defining ankyloglossia: a case series of anterior and posterior tongue ties. *Int J Pediatr Otorhinolaryngol*. 2010 Sep;74(9):1003-6. doi: 10.1016/j.ijporl.2010.05.025. PMID: 20557951.
5. Messner AH, Walsh J, Rosenfeld RM, et al. Clinical Consensus Statement: Ankyloglossia in Children. *Otolaryngol Head Neck Surg*. 2020 May;162(5):597-611. doi: 10.1177/0194599820915457. PMID: 32283998.
6. Thomas J, Bunik M, Holmes A, et al. Identification and Management of Ankyloglossia and Its Effect on Breastfeeding in Infants: Clinical Report. *Pediatrics*. 2024;154(2):e2024067605. doi: 10.1542/peds.2024-067605.
7. Kotlow LA. Ankyloglossia (tongue-tie): a diagnostic and treatment quandary. *Quintessence Int*. 1999 Apr;30(4):259-62. PMID: 10635253.
8. Walker RD, Messing S, Rosen-Carole C, McKenna Benoit M. Defining Tip-Frenulum Length for Ankyloglossia and Its Impact on Breastfeeding: A Prospective Cohort Study. *Breastfeed Med*. 2018 Apr;13(3):204-10. doi: 10.1089/bfm.2017.0116. PMID: 29620937.
9. Brzęcka D, Garbacz M, Micał M, et al. Diagnosis, classification and management of ankyloglossia including its influence on breastfeeding. *Dev Period Med*. 2019;23(1):79-87. doi: 10.34763/devperiodmed.20192301.7985. PMID: 30954985.
10. Hogan M, Westcott C, Griffiths M. Randomized, controlled trial of division of tongue-tie in infants with feeding problems. *J Paediatr Child Health*. 2005 May-Jun;41(5-6):246-50. doi: 10.1111/j.1440-1754.2005.00604.x. PMID: 15953322.
11. Coryllos E, Genna C, Salloum AC. Congenital tongue-tie and its impact on breastfeeding. *American Academy of Pediatrics Section on Breastfeeding*. 2004 01/01:1-6.
12. Kotlow L. Infant Reflux and Aerophagia Associated with the Maxillary Lip-tie and Ankyloglossia (Tongue-tie). *Clinical Lactation*. 2011 12/01;2:25-9. doi: 10.1891/215805311807011467.
13. Geddes DT, Langton DB, Gollow I, et al. Frenulotomy for breastfeeding infants with ankyloglossia: effect on milk removal and sucking mechanism as imaged by ultrasound. *Pediatrics*. 2008 Jul;122(1):e188-94. doi: 10.1542/peds.2007-2553. PMID: 18573859.
14. Aldulaijan HA, Alsharif SB, Cohen RE, Yerke LM. Familial Presentation of Ankyloglossia and Localized Periodontitis. *Compend Contin Educ Dent*. 2022 May;43(5):288-91. PMID: 35589147.

15. Blancher A, Mamidi I, Morris L. Tongue and Mandibular Disorders of the Pediatric Patient. *Facial Plast Surg Clin North Am.* 2024 Feb;32(1):157-67. doi: 10.1016/j.fsc.2023.08.002. PMID: 37981411.
16. Kent JC, Ashton E, Hardwick CM, et al. Nipple Pain in Breastfeeding Mothers: Incidence, Causes and Treatments. *Int J Environ Res Public Health.* 2015 Sep 29;12(10):12247-63. doi: 10.3390/ijerph121012247. PMID: 26426034.
17. Webb AN, Hao W, Hong P. The effect of tongue-tie division on breastfeeding and speech articulation: a systematic review. *Int J Pediatr Otorhinolaryngol.* 2013 May;77(5):635-46. doi: 10.1016/j.ijporl.2013.03.008. PMID: 23537928.
18. Wei EX, Meister KD, Balakrishnan K, et al. Ankyloglossia: Clinical and Sociodemographic Predictors of Diagnosis and Management in the United States, 2004 to 2019. *Otolaryngol Head Neck Surg.* 2023 Oct;169(4):1020-7. doi: 10.1002/ohn.332. PMID: 36994937.
19. UNDP (United Nations Development Programme). Human Development Report 2023-24: Breaking the gridlock: Reimagining cooperation in a polarized world. New York, NY: 2024. <https://hdr.undp.org/content/human-development-report-2023-24>