Boston College

William F. Connell School of Nursing

# TONGUE-TIE: PREVALENCE, SIGNIFICANCE, AND ITS CONTRIBUTION TO MATERNAL FEEDING CHALLENGES AND PROBLEMATIC INFANT FEEDING

a dissertation

by

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#### Abstract

**Background.** Ankyloglossia, commonly known as tongue-tie, reduces tongue mobility through restriction of the lingual frenulum. A recent professional consensus notes that tongue-tie is one possible reason for breastfeeding challenges. Prevalence reporting of tongue-tie has been variable, and an understanding of which infants benefit most from treatment is unknown. Screening and treatment guidelines have not been developed, mainly due to low-level evidence in published research. Despite this, there has been a substantial increase in tongue-tie treatment via frenotomy in the last 15 years.

**Purpose.** The purpose of this program of research was to improve our understanding of tongue- tie and its impact on infant feeding. Eight specific aims were developed to achieve this purpose: 1) identify the prevalence rate of anyloglossia in infants age birth through 12 months; 2) review and evaluate the diagnostic criteria used to diagnose ankyloglossia; 3) identify and summarize original research addressing the impact of ankyloglossia on infant feeding, comparing symptoms of problematic feeding before and after frenotomy; 4) review the quality of the feeding-related outcome measures and psychometric properties of the assessment tools used; 5) describe changes in problematic feeding symptoms, as measured by the NeoEAT, pre- and post-frenotomy; 6) explore the contribution of infant age to the magnitude of change in problematic feeding pre- and post-frenotomy; 7) describe changes in maternal symptoms pre- and post-frenotomy; and 8) evaluate the relationships between maternal symptoms and symptoms of problematic feeding pre- and post-frenotomy.

**Methods.** First, we performed a systematic review and meta-analysis of the literature on tongue- tie to determine the prevalence of the anomaly in the infant population and critiqued the methods used to achieve tongue-tie diagnosis. Second, we identified and summarized

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original research addressing the impact of tongue-tie on infant feeding, comparing symptoms of problematic feeding before and after frenotomy. In this same study, we evaluated the psychometric properties of the assessment tools used in the published research. Next, we assessed maternal and infant symptoms of problematic feeding pre- and post-frenotomy. In this same study, we utilized a comprehensive evaluation of infant feeding symptoms using a validated measure.

Results. Cumulatively, this dissertation research has identified symptoms in both mothers and their infants in the setting of tongue-tie. Each of the eight specific aims proposed for this dissertation were addressed. Specifically, through the meta-analysis and systematic review, we determined 1) prevalence of tongue-tie is higher than previously thought, affecting 8% of the infant population, 2) current screening tools for tongue-tie require psychometric evaluation, 3) LATCH scores and maternal self-efficacy improve following frenotomy but little is known about the effect of frenotomy on infant feeding, and 4) infant feeding has not been evaluated comprehensively or with a validated measure for babies with tongue-tie. The research study conducted in Chapter IV found that 5) infants with severe tongue-tie experienced significant improvements in problematic feeding symptoms following tongue-tie correction as measured by the NeoEAT, 6) regardless of infant age, improvements in symptoms of problematic feeding were seen post-frenotomy in babies with severe tongue-tie, 7) maternal symptoms previously thought to occur in the setting of tongue-tie improved following frenotomy (e.g., painful or difficult latch), and 8) symptoms that have not been assessed in earlier research also improved after tongue-tie correction (e.g., chewing on nipple, incomplete breast drainage, and over supply of breastmilk).

Conclusions. The program of research in this dissertation has made meaningful contributions

to the literature on tongue-tie. Through the production of the first meta-analysis of prevalence data, a thorough evaluation of the available research, and the determination of maternal and infant symptoms associated with tongue-tie, we have crafted recommendations for future research and recommendations for improvements in the clinical management of infants with this oral anomaly.

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I dedicate this dissertation to you, dad. I hope I have made you proud.

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Chapter I: Introduction to the dissertation Rebecca R. Hill, DNP, MSN, FNP-C

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### Introduction

A three-manuscript dissertation has been chosen in lieu of the traditional dissertation option. Chapter I presents an introduction to the program of research proposed for this dissertation, including the following sections: significance of the research proposed; brief review of the literature; overarching theoretical framework; overall purpose of the body of work and associated aims; and implications of the research for nursing practice and science.

### **Statement of the Problem**

Ankyloglossia, commonly known as tongue-tie, is a minor congenital anomaly resulting in a tight connection of the tongue to the floor of the mouth (Messner, Lalakea, Aby, Macmahon, & Bair, 2000). Recent evidence suggests a prevalence rate of tongue-tie between 0.3% (Çetinkaya et al., 2011) to 16% (Ngerncham et al., 2013), but true prevalence of the condition remains unknown. A genetic predisposition has been identified, linked to "a mutated T-box transcription factor gene" (Hazelbaker, 2010, p. 46), with a male predominance of the anomaly.

Tongue-tie has been mentioned in literature dating back to the 1700's (Theobald, 1760). In the 18<sup>th</sup> century, midwives would cut the tight frenulum with a sharp fingernail, and it was routinely treated up through the 1940's (Wright & Waterston, 2006). Around that time, infant formula became more commonplace, the promotion of breastfeeding (BF) by healthcare providers declined, and screening for ankyloglossia decreased (Wright & Waterston, 2006). Health professions education began omitting training on screening and treatment of tonguetie in the 1950's. Over the last half century, benefits of BF for both mother and baby have been identified (Eidelman & Schanler, 2012), with strategies from healthcare organizations aimed at increasing BF rates nationally and internationally. With this interest in increasing BF rates, there has been renewed interest in the contribution of tongue-tie to BF difficulty and cessation.

Exclusive breastfeeding (EBF) is defined by the World Health Organization as receiving only breastmilk, without other liquids or foods, from the breast, bottle, or wet nurse (WHO, 2020). Currently, less than forty percent of all children worldwide are breastfed exclusively for the first six months of life (UNICEF, 2015). These suboptimal rates of EBF contribute to overall childhood mortality in children less than five years of age (Summers & Bilukha, 2018) as well as the occurrence of acute and chronic illnesses in childhood.

The Agency for Healthcare Research and Quality (AHRQ) summarized findings from meta-analyses comparing infant health outcomes between infants fed with breastmilk and those fed with formula ("Systematic Review of Breastfeeding Programs and Policies, Breastfeeding Uptake, and Maternal Health Outcomes in Developed Countries | Effective Health Care Program,"). Risk of common childhood illnesses and serious diseases are more likely in formula-fed infants, including asthma (Oddy, 2017), middle ear infections (Li, Dee, Li, Hoffman, & Grummer-Strawn, 2014), and necrotizing enterocolitis (Herrmann & Carroll, 2014). Risk of chronic diseases is reduced in infants who are breastfed (Yan, Liu, Zhu, Huang, & Wang, 2016). The Centers for Disease Control and Prevention (CDC) report a protective benefit of breastfeeding against obesity, with a longer duration of breastfeeding directly linked to a lower risk of becoming obese (CDC, 2018). Breastfeeding women are less likely to develop ovarian (Li et al., 2014) or breast cancer (Franca-Botelho, Ferreira, Franca, Franca, & Honorio-Franca, 2013). The positive effects associated with breastfeeding transcend all societal groups, regardless of income or status (Jones, Power, Queenan, & Schulkin, 2015).

Improvements in the rates of EBF was a main objective in Healthy People 2020 and appears to remain a goal in the Healthy People 2030 draft (CDC, 2014; CDC, 2019). In 2011, the Surgeon General and United States Department of Health and Human Services published a call to action to promote and support breastfeeding efforts. This call demanded attention to the roles and responsibilities of clinicians, communities, and researchers to "enable mothers to meet their personal goals for breastfeeding" (p. v).

Despite recent initiatives and organizational recommendations, the rate of EBF in the United States remains low. The rate is even lower in racial and ethnic minority populations, where breastfeeding may be of greater benefit given their disproportionately higher rate of adverse health outcomes (Jones, Power, Queenan, & Schulkin, 2015). Data from the Ross Mothers Survey from 1970-1998 demonstrated a steady increase in breastfeeding rates. In 1970, less than ten percent of women were breastfeeding. In 2016, 83.8% of mothers initiated breastfeeding, implying a strong initial desire and interest for women to breastfeed their infants. However, the rates for both BF duration and EBF continue to fall short. The CDC's Breastfeeding Report Card data from 2016 shows that EBF through 6 months occurs for 25.4% of infants; 17.2% of infants received formula before two days of age (CDC, 2016). The imbalances in the rate of BF initiation compared with long-term EBF rates suggests there are barriers following hospital discharge precluding mothers from continuing to breastfeed.

One of the most frequently reported reasons for the cessation of breastfeeding earlier than anticipated is the presence of nipple pain (Kent et al., 2015). While discomfort with feeding can occur for a variety of reasons, tongue-tie is one potential cause and a common reason for mothers seeking treatment for discomfort with feeding (Kotlow, 2004). Infants with tongue-tie are three times more likely to be bottle-fed by one week of age (Ricke, Baker, Madlon-Kay, & DeFor, 2005). This tightened oral tissue restricts the tongue's mobility, making the innate physiologic processes necessary for infant feeding (e.g., sucking, swallowing) difficult.

### Pathophysiology of Tongue-Tie and the Effect on Feeding

The tongue begins to develop in the fourth week of gestation, with the musculature created by myoblasts. The ectoderm is responsible for continued growth of the tongue in a downward fashion, allowing the tongue to separate partially from the floor of the mouth. When the ectoderm fails to degenerate properly, tongue mobility is restricted, resulting in tongue-tie. This has been documented as a failure of apoptosis, or normal programmed cell death that prevents the destruction of excess frenulum tissue (Avery & Chiego, 2019). The embryonic development of the tongue creates two distinct portions: oral and pharyngeal. The oral portion includes the tongue tip, blade, dorsum and sides. The pharyngeal portion is comprised of the root of the tongue, which extends from the circumvallate papillae to the hvoid bone. During the 8<sup>th</sup> week of fetal development, the palate works to push the tongue further down within the oral cavity. The tongue begins to widen, which assists with the medial growth of the palate, expanding the palate when the fetus practices swallowing, and the tongue moves upward. By the 28<sup>th</sup> week of fetal development, mouth movements can be seen on ultrasound and by 40 weeks, the suck-swallow-breathe (SSB) mechanism has matured, and the infant is prepared to begin feeding at birth. The SSB mechanism occurs through a variety of structures, including muscles and nerves, working together to intake air and food, both vital to sustain life.

Specific to the tongue, the glossopharyngeal and hypoglossal cranial nerves represent the main sources of innervation. The glossopharyngeal nerve provides taste sensation for the posterior third of the tongue while the hypoglossal nerve serves only the tongue, controlling motor function of the muscle. The tongue is crucial for successful feeding, responsible for several actions that ensure successful seal and swallowing.

The oral phase of swallowing includes rooting, attachment, and sucking (Genna, 2017). The rooting reflex is a vital component for successful feeding and occurs when the infant reflexively turns toward a stimulus and begins sucking motions to prepare for feeding. Attachment to the breast or bottle, also known as latch, is required for successful feeding. Genna (2017) states "the infant's tongue position during approach to the breast is one of the most important factors in successful attachment" (p. 34). When the infant initially latches, the front portion of the tongue elevates, a mechanism that may be partially or completely impaired with excess frenum tissue.

Sucking occurs after rooting and latching. Here, the tongue forms a lateral groove to move fluid to the back of the throat. Range of motion of the tongue is critical for the oral stages of sucking and swallowing. In physics, Boyle's Law states that movement occurs from high to low pressure areas. This applies to the swallowing of fluid, with the tongue responsible for the initiation and sustainment of this pressure gradient. To ensure optimal pressure changes during sucking, the tongue organ plays a key role in generating a seal of the oral cavity – first through the tongue-tip placement that wraps around the underside of the breast nipple or bottle teat, and secondly by way of the apposition to the soft palate that seals off the posterior cavity during the milk expression phase. As the jaw drops during the milk expression phase, the tongue depresses posteriorly, allowing negative pressure to increase, drawing milk out of the breast or bottle. The tongue also plays a significant role in propelling the bolus into the pharynx, initiating the swallow. A retraction movement of the posterior

tongue, coupled with posterior tongue depression, creates a ramp and a backward thrust of the bolus from the ramped tongue (Hazelbaker, 2010, p. 56). Real-time 3D ultrasonography has revealed that peristaltic movement of the tongue is required for milk transfer while breastfeeding (Burton, Deng, McDonald, & Fewtrell, 2013). When an infant has a tongue-tie, the movement of the tongue may be restricted, resulting in difficulty establishing and maintaining a latch and pressure seal, and difficulty moving the tongue in a manner to successfully and efficiently remove milk from either the breast or bottle. To date, there is no published research that has examined suck qualities in infants with tongue-tie using a valid and reliable measure of this physiologic process.

During swallowing, the vocal folds and epiglottis close to protect the airway and the base of the tongue contacts the posterior pharyngeal wall to create pressure to move the bolus downward (as opposed to moving up into the nasopharynx) (Morris & Klein, 2000). If the movement of the tongue is restricted, it may prevent the tongue from making appropriate contact with the posterior pharyngeal wall, resulting in nasopharyngeal reflux, and may interfere with the closure of the structures involved in protecting the airway, placing the infant at risk for aspiration. Disorganized swallowing in the setting of tongue-tie may allow the infant to swallow excess air, causing aerophagia-induced reflux (Siegel, 2017).

### Symptoms of Feeding Difficulty Associated with Tongue-Tie

The abnormal tongue mechanics that occur with tongue-tie result in the symptoms of feeding difficulty commonly associated with the diagnosis. When compared with infants without tongue-tie, "most are less efficient than their peers" (Geddes et al., 2010). Transfer of milk may be incomplete, resulting in inadequate amounts necessary to sustain growth. Inability to transfer milk directly impacts maternal milk supply, as milk production is

dependent upon emptying the breast.

With impaired tongue movement, the infant may cough, make loud gulping sounds, or ingest excessive air (aerophagia). Excessive gas, reflux, colic and slowed weight gain may manifest in the infant (Kotlow, 2004). The breastfeeding mother may experience frequent clogged ducts, nipple pain, or mastitis (Kotlow, 2004). When the tongue cannot extend below the lower gum line, reflexive biting occurs and the infant may chew, rather than suck, on the nipple (Watson Genna, 2017). This reflex may be responsible for the nipple pain and damage endorsed by mothers breastfeeding infants with tongue-tie. Poor tongue elevation impairs the ability of the palate to widen. When the palate is narrow, a hypersensitive gag reflex results, limiting the infant's ability to accept the breast or bottle nipple deeply. The superficial intake of the nipple creates friction, blistering, and pain of the maternal nipple. While there have been several studies evaluating the effect of frenotomy on infant feeding (Ballard, Auer, & Khoury, 2002; Braccio, Chadderton, Sherridan, & Upadhyaya, 2016; Buryk, Bloom, & Shope, 2011; Dollberg, Marom, & Botzer, 2014; Emond et al., 2014; Geddes et al., 2008; Ghaheri, Cole, Fausel, Chuop, & Mace, 2017; Ghaheri, Cole, & Mace, 2018; Hogan, Westcott, & Griffiths, 2005), none have used a comprehensive measure to evaluate symptoms of problematic feeding. The available research has used measures that focus mostly on maternal symptoms, lack sound psychometric properties, or evaluate only a portion of infant mechanics (i.e., audible swallowing). Thus, the lack of comprehensive infant evaluation in the setting of tongue-tie and following frenotomy is a major gap in the published research.

Frequently, upper lip tie (ULT) is diagnosed concurrently with tongue-tie, and there are postulations that a restricted upper lip can negatively impact feeding from impaired flanging of the lip and improper suction necessary for feeding (Nakhash, Wasserteil, Mimouni, Kasirer, Hammerman, & Bin-Nin, 2019). When the upper lip is improperly flanged and overused, sucking blisters develop (Genna, 2017). It is important to note that consensus agreement by providers on the diagnosis of lip tie has not been agreed upon, nor has it been determined to be a cause of breastfeeding problems (Messner et al., 2020).

### **Assessment of Tongue-Tie**

In the United States, there are no guidelines for the assessment or management of tongue- tie. This mild congenital anomaly is diagnosed by visualization of the anatomy and function of the tongue. Assessment of the tongue may include observations of tongue extension, lateralization, and elevation. Normally, the tongue should protrude below the lower gum line, move equally side-to-side, and the tip should rise to the palate (Genna, 2017). In the presence of tongue-tie, the range of motion is impaired, directly impacting extension, lateralization, and elevation. Tongue-tie can be classified as anterior or posterior and there are several different classification systems to grade its severity. Most often, the anterior tongue-tie is visible on oral examination and visualization of the posterior tongue-tie requires greater tongue elevation to diagnose. The available literature largely focuses on anterior tongue-tie. One screening instrument developed by an expert in the field recommends classifying ankyloglossia based on the distance of the frenulum attachment to the tip of the tongue, with measurements in millimeters equating with mild, moderate, severe, or complete ankyloglossia (Kotlow, 2004).

Beyond the length of the freely mobile tongue, the clinician should be trained to examine the oral cavity in a systematic fashion, using inspection and palpation to identify the presence of a tight frenulum. First, ability to extend the tongue beyond the bottom lip is examined. Next, the Murphy maneuver is used. With the fifth digit, a sweeping motion is completed under the tongue from one side to the other, palpating for a "speed bump" which could indicate the presence of a tight frenulum (Manipon, 2016). Other screening tools incorporate the tongue mechanics of elevation, lateralization, and cupping (Hazelbaker, 2010). None of the published screening tools have been validated for universal use.

The diagnosis of tongue-tie is frequently missed at birth and treatment is delayed, due in part to lack of validated screening measures (Siegel, 2017). The use of a valid and reliable measure is critical to ensure an accurate assessment of the oral cavity, proper diagnosis of the anomaly, and provision of inter-rater consistency. In Brazil, a neonatal assessment for tongue-tie has been adopted as a standard of care in 2014 under law 13.002. This law states that "all newborns in the country must undergo lingual frenulum assessment using the Lingual Frenulum Protocol for Infants (LFPI) – Neonatal Tongue Screening Test before hospital discharge" (de Castro Martinelli et al., 2016, p. 1324). Additional research is needed before standards of care can be implemented in the United States. The published research cannot be used to develop screening or treatment guidelines because of the varied methods used to diagnose and classify tongue-tie across studies.

### **Treatment of Tongue-Tie**

Current practice for the treatment of tongue-tie is not standardized and insurance coverage for treatment is variable (Walsh & Tunkel, 2017). While many providers trained in frenotomy recommend every case of tongue-tie is corrected, a great deal of controversy remains within the medical community and parents with regard to whether or not this recommendation has resulted in over-treatment for the subset of the population that has gained increased awareness of the anomaly. A recent study reported an 866 percent increase in frenotomy rates from 1997-2012 (Walsh, Links, Boss, & Tunkel, 2017). There is a dichotomy of opinions between providers that specialize in infant feeding and those who consider the diagnosis of tongue-tie to be a fad and money-making scheme by providers performing revisions of the anomaly, as lack of insurance coverage frequently results in out-of-pocket payment by families (Walsh, Links, Boss, & Tunkel, 2017).

There are a variety of intervention techniques to treat tongue-tie. Frenotomy can be performed with or without local anesthetic and the tongue-tie can be released using scissors, a scalpel, or with electrocautery or diode laser (Ghaheri et al., 2017; Ramoser et al., 2019). The technique used is provider and training dependent. A recent systematic review found that current literature does not demonstrate advantages for any specific frenotomy technique (Khan, MacPherson, Bezuhly, & Hong, 2020). Frequently, upper lip ties are treated in the same visit. The procedure does not require general anesthesia. Risks of frenotomy include bleeding, infection, or incomplete release of the restriction, requiring additional treatment. Severe or life-threatening complications have not been documented. Following the procedure, postprocedural stretches are typically advised to avoid regrowth of the frenulum tissue. These stretches involve gentle lifting of the upper lip and tongue every four to six hours for at least two weeks post revision (Ghaheri et al., 2017). However, a recent consensus statement found that there is no evidence to support stretches or any other post-care regimen (Messner et al., 2020). In 2017, a Cochrane review determined that there is insufficient evidence to recommend frenotomy for tongue-tie. The studies reviewed had small sample sizes and methodological flaws. Despite short-term decreases in maternal nipple pain, long-term improvements and infant outcomes were not addressed to support frenotomy (O Shea et al., 2017).

### **Theoretical Framework**

Using the Theory of Unpleasant Symptoms (Lenz, Pugh, Milligan, Gift, & Suppe, 1997) as the guiding theoretical framework in this dissertation (Figure 1), a deeper understanding of tongue-tie as it relates to the health and well-being of infants will be achieved. The Theory of Unpleasant Symptoms (Lenz, Pugh, Milligan, Gift, & Suppe, 1997) is a middle-range theory designed to integrate the existing knowledge of symptoms that may guide future research and practice. The major tenets of this theory include the experience of symptoms, influencing factors that promote or alleviate symptoms, and the consequences associated with the symptoms (Lenz, Pugh, Milligan, Gift, & Suppe, 1997). This theory and its conceptual model emphasize three main factors that contribute to the symptom experience: physiologic, psychologic, and situational. The culmination of these factors creates the perceived symptom(s) which in turn affects performance. Infant feeding is the performance activity of interest, examined through a variety of lenses in each of the proposed dissertation manuscripts.

Factors may be present in both the mother and the infant, resulting in a wide array of symptoms that influence infant feeding. This program of research will focus on the infant symptoms of feeding difficulty associated with tongue-tie, the influencing factors that promote or alleviate symptoms, including frenotomy, and the consequences to the infant because of those symptoms. A critical examination of the methods used to diagnose tongue-tie and the reported prevalence rates across studies will be evaluated. The systematic review and meta-analysis will be informed by this middle-range theory. Often, the diagnosis of tongue-tie is based on functional capabilities of the infant's tongue and its mobility, which directly relates to symptoms associated with tongue-tie and the infant's ability to successfully feed from a nipple or bottle teat. These physiologic mechanisms are evaluated using

classification systems such as the Coryllos, Kotlow or Hazelbaker assessments of lingual function. The first manuscript proposed for this dissertation will analyze these assessment measures, compare them across studies, and evaluate their utility to diagnose tongue-tie on the basis of maternal and/or infant symptoms (Chapter II).

The systematic review and examination of the psychometric properties of the tools used to evaluate feeding will be examined in the second manuscript (Chapter 3). The evaluation of the symptoms reported prior to and following frenotomy will be critically appraised to develop a clear understanding of the current state of the science and identify gaps in the research that necessitate future study. The gaps will be framed from the perspective of unpleasant symptoms associated with tongue-tie. Finally, the research study proposed for this dissertation will integrate the information from the first two manuscripts to conduct a study that not only utilizes comprehensive, valid and reliable measures of infant feeding, but also addresses critical gaps in the field of tongue-tie and frenotomy. The Theory of Unpleasant Symptoms will serve as the context for designing the research study, to include the basic tenants of symptom recognition and assessment and compare those unpleasant symptoms before and after correction of tongue-tie via frenotomy. The goal of symptom evaluation in this dissertation will be to better understand how tongue-tie and its correction impact the performance activity of interest, infant feeding.

### Purpose/Specific Aims

The overall purpose of this body of work is to improve our understanding of tongue-tie and its impact on infant feeding. The known gaps in the literature (i.e., true prevalence, effects of frenotomy on feeding, and absence of comprehensive infant feeding measures) will be addressed in this body of work through the production of three manuscripts. Specific aims have been identified and set forth for this dissertation (Table 1). These aims represent the work of a meta- analysis, systematic review, and original research study that together, will increase our understanding of tongue-tie and the effectiveness of frenotomy.

The specific aims for this body of work are: (see Table 1 for details on which chapter will address each aim):

Aim 1: Identify the prevalence rate of ankyloglossia in infants age birth through 12 months.

Aim 2: Review and evaluate the diagnostic criteria used to diagnose ankyloglossia.

**Aim 3:** Identify and summarize original research addressing the impact of ankyloglossia on infant feeding, comparing symptoms of problematic feeding before and after frenotomy.

**Aim 4:** Review the quality of the feeding-related outcome measures and psychometric properties of the assessment tools used.

**Aim 5:** Describe changes in problematic feeding symptoms, as measured by the NeoEAT, pre- and post-frenotomy.

**Aim 6:** Explore the contribution of infant age to the magnitude of change in problematic feeding pre- and post-frenotomy.

Aim 7: Describe changes in maternal symptoms pre- and post- frenotomy.

**Aim 8:** Evaluate the relationships between maternal symptoms and symptoms of problematic feeding pre- and post-frenotomy.

Specific Aim	Chapter / Title of Paper
Aim #1: Identify the prevalence rate of ankyloglossia in infants age birth through 12 months Aim #2: Review and evaluate the diagnostic criteria used to diagnose ankyloglossia	Chapter 2: The prevalence of ankyloglossia in infants: a systematic review and meta- analysis
Aim #3: Identify and summarize original research addressing the impact of ankyloglossia on infant feeding, comparing symptoms of problematic feeding before and after frenotomy.	Chapter 3: Symptoms of problematic feeding in infants under 1 year of age undergoing frenotomy: A review article
Aim #4: Review the quality of the feeding-related outcome measures and psychometric properties of the assessment tools used.	
Aim #5: Describe changes in problematic feeding symptoms, as measured by the NeoEAT, pre-and post-frenotomy.	Chapter 4: The influence of tongue-tie and frenotomy on maternal breastfeeding
Hypothesis 1: NeoEAT total and subscale scores will	symptoms and problematic infant feeding
decrease after frenotomy.	
Aim #6: Explore the contribution of infant age to the magnitude of change in problematic feeding pre- and post-frenotomy.	
<i>Hypothesis 1:</i> Infant age at time of frenotomy will have a statistically significant relationship with magnitude of change, with younger infants showing greater improvement in problematic feeding post-frenotomy.	
Aim #7: Describe changes in maternal symptoms pre- and post-frenotomy.	
<i>Hypothesis 1:</i> Breastfeeding mothers will report a decrease in maternal symptoms from pre- to post-frenotomy.	
Aim #8: Evaluate the relationships between maternal symptoms and symptoms of problematic feeding pre- and post-frenotomy.	
<i>Hypothesis 1:</i> More severe maternal symptoms (e.g., bleeding nipples, painful latch, abraded nipples) will be highly correlated with higher NeoEAT scores both pre- and post-frenotomy.	

### Table 1: Outline of Specific Aims and Papers to Address Aim(s)

### Implications of the Research for Nursing Science

The cumulative research proposed for this dissertation will contribute substantially to

### Rebecca Hill: Manuscript Style Dissertation

nursing science and practice. First, through the exploration on the current literature, clear gaps in the state of the science will be identified to drive the research study proposed to meet aims five, six, seven, and eight. The analysis of outcome measures used to evaluate feeding will be critiqued to provide rationale for use in the development of the original research study proposed. The strengths and limitations discovered in the systematic review of the literature will assist with the development of specific aims, inclusion and exclusion criteria and variables to account for when planning the research study. The meta-analysis and synthesis of current diagnostic criteria will provide a better understanding of the prevalence of ankyloglossia while also guiding the research and diagnostic criteria that will be used in the research study. Following completion of aims five through eight, there will be contributions to nursing science that will provide the groundwork for a longitudinal program of research. This research will offer the foundation toward development of screening, diagnostic, and treatment guidelines that will span a multitude of healthcare disciplines with the goal of improved care for women and infants.

#### Summary

The program of research in this proposed dissertation will address critical areas in the care of infants with tongue-tie, its impact on feeding, and the role of frenotomy. The systematic review and meta-analysis will provide greater understanding of the true prevalence rate of ankyloglossia, while concurrently evaluating the varied diagnostic criteria being used nationally and internationally. The synthesis of the current evidence for frenotomy in infants with tongue- tie is an initial step in understanding the state of the science, gaps in the research, and the implications and limitations of the published work to date. Finally, several of these gaps and limitations will be addressed in a prospective study using a standardized measure for

diagnosis and valid, reliable tools to evaluate problematic infant feeding. The cumulative body of work that will be a product of this dissertation is the first step toward improved understanding of the effect of tongue-tie on infant feeding and the role frenotomy may have in alleviation of problematic feeding in this population to support breastfeeding and overall health.





### References

### **Refer to Cumulative Reference List**

### Chapter II

The prevalence of ankyloglossia in children aged <1 year: a systematic review and meta-analysis Rebecca R. Hill, DNP, MSN, FNP-C Boston College William F. Connell School of Nursing Christopher Lee, PhD, RN, FAHA, FAAN, FHFSA Boston College William F. Connell School of Nursing Britt F. Pados, PhD, RN, NNP-BC, CLC, FAHA, FNAP Boston College William F. Connell School of Nursing

This manuscript replaces portions of the methods section and results section of the traditional dissertation. Rebecca Hill was the primary author on this paper, and Dr. Pados was the senior author. Rebecca Hill completed the systematic review, meta-analysis, and manuscript preparation under the supervision of Dr. Pados. Dr. Lee was the second author and provided expertise on the statistical methods to conduct the meta-analysis. The readership for this journal includes healthcare professionals, particularly providers of care such as physicians, nurse practitioners, and physician assistants. This manuscript was published on November 13, 2020 by Pediatric Research, which has an impact factor of 2.880. This manuscript represents a significant contribution to the dissertation work.

### Abstract

Background: Despite the low-level of evidence supporting the correction of tongue-tie for breastfeeding problems, recognition and treatment has increased substantially over the last 15 years. Prevalence reporting of tongue-tie is variable. The purpose of this study was to quantitatively synthesize the prevalence of tongue-tie in children under one year of age and to examine the psychometric properties of the assessment tools used for diagnosing tongue-tie in these studies.

Methods: PRISMA and MOOSE guidelines were followed, with selection of studies and data extraction verified by two authors. Random-effects meta-analyses were performed to determine an overall prevalence rate, prevalence by infant sex, and prevalence by diagnostic method.

Results: There were 15 studies that met inclusion criteria. Overall prevalence of tongue-tie (N = 24,536) was 8% (95% CI 6 – 10%, p < 0.01). Prevalence was 7% in males and 4% in females. Prevalence was 10% when using a standardized assessment tool compared to 7% when using visual examination alone (p = 0.16). Available assessment tools for diagnosis of tongue-tie do not have adequate psychometric properties.

Conclusion: Tongue-tie is a common anomaly, which has the potential to impact infant feeding. Development of a psychometrically-sound assessment of tongue-tie is needed.

Keywords: Ankyloglossia; Tongue-tie; Prevalence

The prevalence of ankyloglossia in children aged <1 year: a systematic review and meta-

analysis

#### Introduction

Ankyloglossia, commonly known as tongue-tie, is an anatomic variation that restricts tongue mobility caused by a restrictive lingual frenulum (Messner et al., 2020), best described as a midline fold (Mills, Pransky, Geddes, & Mirjalili, 2019). The structure of the lingual frenulum consists of a "dynamic three-dimensional structure that varies in morphology on a spectrum" (Mills et al., 2019, p. 760). This anatomical variation of the lingual frenulum can be classified by height of the facial attachment to the ventral portion of the tongue, height of facial attachment to the tongue, height of attachment to the mandible, and length of the frenulum (Mills et al., 2019). This variability is classified by how the fibers of the lingual frenulum mobilize when the fascia is placed under tension (Mills et al., 2019). The restricted motion of the tongue can alter oral latch onto a breast or bottle nipple and impact swallowing, which may cause ineffective transfer of breastmilk or formula (Geddes et al., 2008). Improper latch to the breast or bottle can impact the infant's ability to obtain adequate nutrition for growth and development and, during breastfeeding (BF), can cause maternal nipple pain and result in early cessation of breastfeeding (Ricke et al., 2005). Prevalence of tongue-tie is not well understood and has been cited as low as 0.3% (Cetinkaya et al., 2011) to as high as 16% (Ngerncham et al., 2013).

Professional consensus on the definition of tongue-tie and how it relates to symptoms of problematic feeding has been developed, noting that ankyloglossia is one possible reason for breastfeeding pain and ineffective latch (Messner & Lalakea, 2000). However, the use of the terms "posterior ankyloglossia" and "lip tie" have not reached professional consensus. In addition, the determination of which infants benefit most from frenotomy to correct tonguetie remains unknown, largely due to the low-level of evidence available, the variability in screening for tongue-tie, and the absence of a psychometrically-sound diagnostic tool (O Shea et al., 2017). Despite the lack of strong evidence, recognition of tongue-tie has increased, with a dramatic rise in treatment via frenotomy of 866% over the last two decades (Walsh & Tunkel, 2017). The purpose of this study was to quantitatively synthesize the prevalence of tongue-tie in children under one year of age and to examine the psychometric properties of the assessment tools used for diagnosing tongue-tie in these studies.

#### Methods

In February 2020, a literature search was conducted to review the published prevalence rates of tongue-tie. Databases searched include the Cumulative Index of Nursing and Allied Health Literature (CINAHL) and PubMed. The search terms ((tongue-tie) or (tongue tie) or (ankyloglossia)) and ((prevalence) or (incidence)) were entered into each database. The search was limited to articles involving humans and published in the English language with a study population of children less than one year of age. No limit was placed on the time of publication. Duplicate articles were removed.

### **Study Selection and Data Extraction**

To be included, articles must have been original research, reporting the prevalence of tongue-tie by screening a larger population. Articles were excluded if they did not include a prevalence rate, if the sample was only a subset of the population that already had a diagnosis of tongue-tie, if participants were over age one, or if the study did not directly assess the sample for tongue-tie (e.g., a chart review or examination of ICD-10 diagnosis codes). Titles and abstracts were screened independently by two authors (R.H. and B.P.) using the inclusion

and exclusion criteria. Articles that met inclusion and exclusion criteria or could not be evaluated based on the abstract were reviewed in full text. Both authors then reviewed the full-text articles independently. Any article that one author chose to include were reviewed by both authors for final decision.

The individual study characteristics were examined in the studies that met criteria for inclusion. The relevant features extracted included the study sample size and characteristics, region of the world where the study took place, diagnostic method used to identify tongue-tie, and reported prevalence of tongue-tie within the sample. Prevalence reported by sex was extracted, if available. The authors conducted this meta-analysis in concordance with Preferred Reporting Items of Systematic Review and Meta-Analyses (PRISMA) standards of quality for reporting meta-analyses (Moher, Liberati, Tetzlaff, & Altman, 2009) and the guidelines for Meta-Analysis and Systematic Reviews of Observational Studies (Stroup, et al., 2000).

### **Examination of Psychometric Properties of Diagnostic Assessments**

Following identification of the diagnostic tools used to identify tongue-tie, a review of the literature was conducted for each of the assessment tools to identify and review their psychometric properties. A search was conducted in both CINAHL and PubMed using the name of each tool separately (Coryllos, Kotlow and the Assessment Tool for Lingual Frenulum Function) and ((psychometrics) or (reliability) or (validity) or (psychometric properties)). This search was limited to humans and English language. Articles reporting the psychometric properties of the assessment tools were examined to review the quality of the assessments.

### **Data Analysis**

An overall prevalence of tongue-tie across studies was examined using a randomeffects meta-analysis of proportions approach. Prevalence was represented as a binomial distribution (tongue-tie present or absent). Subgroup meta-analysis was performed to examine the influence of sex (male or female) and diagnostic method on the prevalence of tongue-tie with studies as the unit of analysis. Random effects tests of difference were then used to compare prevalence between studies of male and female infants, and studies diagnosing tongue-tie by visual exam only compared with those using visual exam in addition to a standardized diagnostic measure.

For all analyses, the use of the random-effects model allowed examination of heterogeneity in and between studies, the appropriate method given that diagnosis is variable and may be subjective. Weighted estimates, 95% confidence intervals (CI), *z*-tests (i.e., summary estimate divided by standard error of the summary estimate), and *p*-values were calculated using Stata v16 (College Station, TX) and the **metaprop** command. Dispersion in effect size across studies (Q) along with an associated p-value, and variation in observed estimates attributable to heterogeneity ( $I^2$ ) were calculated. An alpha of .05 was considered statistically significant.

#### **Assessment of Risk of Bias**

Risk of bias in measurement was assessed through the evaluation of the psychometric properties of the assessment tools used to diagnose tongue-tie. The psychometric properties were extracted by one author (R.H.) and evaluated by two authors (R.H. and B.P.). Risk of bias in prevalence reporting was evaluated using an existing tool to examine external and internal validity and overall risk of bias (Hoy et al., 2012). Overall bias was recorded as low, moderate, or high based upon the 10 items critiqued within each study.

#### Results

The selection process for the literature included in this review is presented in the PRISMA flow diagram (Figure 1). After screening titles and abstracts, 31 full-text articles were reviewed and 15 were eligible for final inclusion in analyses. Included studies were conducted between 1990 and 2019 throughout the world, including in the United States, Thailand, Mexico, Spain, Turkey, Sweden and the United Kingdom. Sample size ranged from 400 (De Oliveira, Duarte, & Diniz, 2019) to 3490 (Ricke et al., 2005). Table 1 presents relevant characteristics extracted from the included studies.

### **Overall Prevalence of Tongue-Tie**

There were 15 studies reporting prevalence of tongue-tie, for a collective total of 24,536 infants (Figure 2). Irrespective of diagnostic method, the overall prevalence was 8% (95% CI 6- 10%, z = 6.51, p = .00). There was significant (Q = 1617.37) and substantial ( $I^2 = 99.13\%$ ) heterogeneity across studies reporting prevalence of tongue-tie.

### **Prevalence by Infant Sex**

Twelve studies reported infant sex. Of those, 8 specifically compared prevalence between female and male infants. Four found a significantly higher incidence of tongue-tie in males (Flinck, Paludan, Matsson, Holm, & Axelsson, 1994; Freudenberger, Santos Díaz, Bravo, & Sedano, 2008; Friend, Harris, Mincer, Fong, & Carruth, 1990; Harris, Friend, & Tolley, 1992) and 4 did not report a significant difference in rates by sex (De Oliveira et al., 2019; Hogan, Westcott, & Griffiths, 2005; Perez-Aguirre et al., 2018; Ricke et al., 2005). Of the 8 studies comparing incidence by sex, 3 were omitted from the analysis because a breakdown of distribution by sex within the full study sample was not provided, resulting in an inability to determine prevalence by sex in those studies (Harris et al., 1992; Hogan, Westcott, & Griffiths, 2005; Ricke et al., 2005). Of the 5 analyzed (De Oliveira et al., 2019; Flinck et al., 1994; Freudenberger et al., 2008; Friend et al., 1990; Perez-Aguirre et al., 2018) (n = 6319), prevalence of tongue was 7% in males (95% CI 2 – 12%, Q = 123.9) and 4% in females (95% CI 1 – 6%, Q = 76.35). The difference in prevalence by infant sex was not statistically significant.

### **Prevalence by Diagnostic Method**

Prevalence of tongue-tie in the ten studies using visual examination alone (n = 14,426) was 7% (95% CI 4 – 10 %, z = 4.47, p = .00, Figure 3). The five studies using a more formal diagnostic method (n = 10,110) had a higher tongue-tie prevalence of 10% (95% CI 6 – 15%, z = 4.83, p = .00). The difference between these sub-groups was not statistically significant (Q = 1.94, p = .16). Substantial variation remained across studies (visual examination method – Q = 1069.58,  $I^2 = 99.16$ %, and standardized measure method – Q = 269.83 Q =,  $I^2 =$ 98.52%).

### **Psychometric Properties of Diagnostic Assessments**

Five studies diagnosed tongue-tie using a published assessment tool, either the Hazelbaker Assessment Tool for Lingual Frenulum Function (ATLFF) (Perez-Aguirre et al., 2018; Ricke et al., 2005), Kotlow's grading system (Puapornpong, Paritakul, Suksamarnwong, Srisuwan, & Ketsuwan, 2017; Puapornpong, Raungrongmorakot, Mahasitthiwat, & Ketsuwan, 2014), or Coryllos classification of tongue-tie severity (Ferrés-Amat et al., 2017). The majority of studies (n = 10) diagnosed based on visual examination, with variation in what criteria supported a diagnosis of tongue-tie. The ability of the infant to protrude the tongue out of the mouth was assessed in three studies (Cetinkaya et al., 2011; Flinck et al., 1994; Friend et al., 1990) and length of frenulum attachment was measured in five studies without specific length necessary for diagnosis explicated (Flinck et al., 1994; Harris et al., 1992; Hogan, Westcott, & Griffiths, 2005; Messner, Lalakea, Aby, Macmahon, Bair, et al., 2000; Ngerncham et al., 2013). Two studies did not explicate criteria used for diagnosis (De Oliveira et al., 2019; Perez-Aguirre et al., 2018).

The ATLFF is a 12-item scale, with five items evaluating the appearance of the tongue and seven evaluating tongue function. Scores range from 0-10 for appearance and 0-14 for function, with higher scores indicating more optimal appearance and function. Hazelbaker recommends a diagnosis of tongue-tie for appearance scores  $\leq 8$  or function scores  $\leq 11$  (A.K. Hazelbaker, 1993). Kotlow's classification system requires the measurement of free tongue in millimeters, classifying tongue-tie severity of Class I through IV based on millimeters of free tongue (Kotlow, 1999). The Coryllos classification system defines four types of frenula based on the location of tongue anchoring. Coryllos recommends this evaluation be done alongside a breastfeeding assessment prior to making the decision to treat (Coryllos, Genna & Salloum, 2004). Of these three tools, only one has published psychometric testing. The ATLFF has demonstrated moderate to excellent inter-rater agreement and correlated with the Bristol Breastfeeding Assessment Tool (Amir, James, & Donath, 2006; Madlon-Kay, Ricke, Baker & Defor, 2008; Ingram, Johnson, Copeland, Churchill, & Taylor, 2015); however, function scores do not correlate with the number of mother-infant dyads who presented with breastfeeding problems caused by tongue-tie (Ricke et al., 2005). The psychometric properties available on these tools are outlined on Table 3.

### **Risk of Bias**

Risk of bias was noted to be moderate or high for all included studies, indicating that future research is likely to affect our confidence in prevalence estimates. While data was collected directly from subjects in all studies, sampling was not random, and it is impossible
to determine if the samples were representative of the population. The definition of tongue-tie differed between studies, interrater reliability was not reported, and there is no psychometric data on the instruments used to measure tongue-tie. Four of the studies failed to include a definition for tongue-tie, resulting in an inability to evaluate exactly what criteria were used to make a diagnosis. The remaining studies included a case definition; however, the studies were conducted prior to the development of professional consensus on the definition of tongue-tie, and uncertainty remains on the threshold of tongue-tie severity warranting treatment via frenotomy. This information is summarized on Table 2.

Egger's test of bias interjected by small studies was significant, indicating that smaller studies reported larger estimates of the prevalence of tongue-tie ( $\beta$ =13.2±2.3; t=5.76; *p* <0.001). A funnel plot (Figure 4) to evaluate possible publication bias did not demonstrate substantive asymmetry. Using meta-regression, there was no significance of year of study publication on the prevalence of tongue-tie reported in the literature ( $\beta$ =0.002±0.001; *t* = 2.02; *p* = 0.065, Figure 5).

## Discussion

This meta-analysis of data from 24,536 infants found the overall prevalence of tongue-tie is 8% in children under 1 year of age. The estimated predictive interval for overall prevalence suggests that future studies may expect to find a prevalence between 0% and 18% using similar approaches to those used in the included studies. Research by Mills and colleagues recognizes that the wide degree of variation in morphology limits our ability to determine a definitive classification for the diagnosis of abnormal frenulum anatomy (Messner et al., 2020). This may partly explain why prevalence of tongue-tie is variable between studies.

A genetic predisposition for tongue-tie has been identified, linked to "a mutated T-box transcription factor gene" (Madlon-Kay, Ricke, Baker & Defor, 2008, p. 46). The higher prevalence found in infants of male compared to female sex is consistent with the majority of studies published to date. The lack of statistical significance of this finding was likely due to the relatively small sample size in the subset of studies reporting prevalence by sex in conjunction with the high degree of heterogeneity in and between studies.

Although not statistically significant, the finding that prevalence of tongue-tie was numerically higher (10%) when a standardized assessment tool is used as compared to visual examination alone (7%) is an important finding. The main limitation in these analyses was that most of the studies used subjective visual examination to diagnose tongue-tie. Of the studies that used a standardized diagnostic tool, the psychometric properties of those tools are not adequate, or have not been evaluated to date. The significant and substantive heterogeneity within and across studies is most likely due to poor measurement of the problem and indicates that differences are due to additional factors beyond the diagnostic method used. Direct comparisons of diagnostic evaluation studies are necessary to understand the clinical importance of the diagnostic method within the same study, not just by comparing studies with different measurement approaches.

Assessment of the quality of tongue-tie assessment tools includes reviewing reliability and validity of the measure. Using measures without strong psychometric properties may not appropriately or accurately measure the outcome variables of interest. Evidence of validity would support that the measure accurately diagnoses tongue-tie, and not a related, but different construct (i.e., normal anatomical variation). Reliability supports that the score is appropriately and reasonably measuring tongue-tie severity and is the same over time and between alternate scorers. Failure to use a psychometrically-sound instrument increases the likelihood that the problem under investigation is not accurately being measured. In this instance, the variation of prevalence rates of tongue-tie between studies may be partly due to the lack psychometric testing and utility of the ATLFF, Coryllos and Kotlow instruments. It is also postulated that in studies using visual examination alone, variability in cut-off scores and what constitutes a diagnosis of tongue-tie contributes to significant heterogeneity. These findings support the need for psychometric evaluation of available assessment tools, or the development of a new, psychometrically-sound, comprehensive tool to diagnose tongue-tie.

One universal diagnostic measure with accepted cutoff scores remains a major limitation in the published research on tongue-tie. Despite lack of psychometric evaluation, the use of any standardized assessment tool to diagnose tongue-tie resulted in an increased prevalence rate when compared to subjective visual examination alone. The ATLFF is the only diagnostic measure that has be evaluated psychometrically. The lack of psychometric evaluation limits the utility of the Kotlow and Coryllos scales to diagnose tongue-tie and future research is needed to assess these measures.

Understanding that tongue-tie diagnosis alone may not constitute difficulty feeding or maternal discomfort with feeding, a comprehensive, objective, and psychometrically-sound evaluation of both infant feeding and maternal symptoms will help determine how tongue-tie impacts infant feeding, maternal comfort and breastfeeding success. The consensus statement by Messner and colleagues was developed solely by otolaryngologists (Messner et al., 2020). More work is needed to incorporate the experience from other experts in the field of infant feeding, including lactation consultants, dentists, and speech and language practitioners. The creation of a diagnostic measure, taking into account the newly revised definition of tonguetie and an interprofessional practice approach, is needed to more accurately capture the prevalence of tongue-tie and reduce heterogeneity between studies. Overall, the study of tongue-tie in infants has been limited by the absence of a universally accepted definition, lack of valid and reliable assessment measures, and the absence of comprehensive evaluation of infant feeding and maternal symptoms. Once these limitations are addressed, a large study is needed to determine the true prevalence of tongue-tie in infants and factors that may be associated with this anomaly.

First Author, Year	Country	Sample Size	Overall Prevalence (%)	Female Prevalence	Male Prevalence	Diagnostic Method
Ballard, 2002	USA	3036	4.2			ATLFF
Cetinkaya, 2011	Turkey	2021	0.3			Visual exam
de Oliveira, 2019	Brazil	400	9	6.8	11.7	Visual exam
Ferres-Amat, 2017	Spain	1102	15.5			Coryllos
Flinck, 1994	Sweden	1021	2.5	1	4.1	Visual exam
Freudenberger, 2008	Mexico	2182	10.6	8.6	12.5	Visual exam
Friend, 1990	USA	500	4.4	2.2	5.7	Visual exam
Harris, 1992*	USA	500	4.4			Visual exam
Hogan, 2005*	UK	1866	10.7			Visual exam
Messner, 2000	USA	1041	4.8			Visual exam
Ngerncham, 2013	Thailand	2679	19.2			Visual exam
Perez-Aguirre, 2018	Mexico	2216	1.5	1.4	1.5	Visual exam
Puapornpong, 2014	Thailand	833	13.4			Kotlow
Puapornpong, 2017	Thailand	1649	14.9			Kotlow
Ricke, 2005*	USA	3490	4.2			ATLFF

# Table 1. Study Characteristics

\*These studies did not report sex breakdown of the full sample and were not included in the prevalence analysis by sex.

Tool (Study)	Intended	Target	Items: Number and	Reliability	Validity
	User	Population	Constructs		
Assessment Tool	Providers	Age < 1	12 items total	Correlated with Bristol	Not tested
for Lingual			Tongue appearance – 5 items	Breastfeeding Assessment	
Frenulum Function			Tongue function – 7 items	Tool ( $r = 0.89, p < .001;$	
(ATLFF)				Ingram et al., 2015).	
(Hazelbaker, 1993)			Scores equate to "perfect"		
			"acceptable" or "function	Moderate reliability of	
			impaired"	"appearance" items (kappa =	
				0.4-0.6), substantial	
			Diagnosis made for	agreement on first 3	
			appearance score $\leq 8$ or	"function" items (kappa =	
			function score $\leq 11$	0.65), "function items related	
				to infant sucking (-0.02-0.05)	
			Score range 0-24, higher	(Amir, 2006).	
			scores indicate better		
			appearance and function	Correlation between function	
				and appearance scores ( $r =$	
				0.49, <i>p</i> < .001) (Ballard,	
				2002).	
				Moderate Inter-rater	
				agreement (kappa = $0.44$ )	
				(Madlon-Kay, 2006)	
				Excellent inter-rater	
				agreement (kappa = 0.92)	
				(Amir, 2006)	

Table 2:	Psychom	etric Pro	perties of	f Assessment	Tools
	2				

				Moderate inter-rater agreement (kappa = 0.49) (Ricke, 2005)	
Kotlow Ankyloglossia Diagnostic Criteria (Kotlow, 1999)	Providers	Designed through examination of children up to age 14	Measurement of free tongue in millimeters (mm) Normal > 16mm Class I (mild) 12-16mm Class II (moderate) 8-11mm Class III (severe) 3-7mm Class IV (complete) < 3mm Plus, structural guidelines to determine if revision is necessary	Not tested	Not tested
Coryllos Classification (Coryllos, 2004)	Providers	Not specifie d	Defines 4 types of frenulum Type 1: tongue anchored from tip, heart-shaped Type II: tongue anchored 2- 4mm from tip Type III: tongue anchored from middle of tongue Type IV: cannot be seen, anchored from base of tongue Recommended for use in conjunction with breastfeeding	No correlation between Coryllos class and breastfeeding difficulties (Haham, 2014)	Not tested

External Validity First Author,	Representative of national population?	Sampling representative of target population?	Random selection of study population?	Minimal non- response bias?
Ballard, 2002	No	No	No	Yes
Cetinkaya, 2011	No	No	No	Yes
de Oliveira, 2019	No	No	No	Yes
Ferres-Amat, 2017	No	No	No	Yes
Flinck, 1994	No	No	No	Yes
Freudenberger, 2008	No	No	No	Yes
Friend, 1990	No	No	No	Yes
Harris, 1992	No	No	No	Yes
Hogan, 2005	No	No	No	Yes
Messner, 2000	No	No	No	Yes
Ngerncham, 2013	No	No	No	Yes
Perez-Aguirre, 2018	No	No	No	Yes
Puapornpong, 2014	No	No	No	Yes
Puapornpong, 2017	No	No	No	Yes
Ricke, 2005	No	No	No	Yes

Internal Validity	Were data collected directly from subjects?	Was an acceptable case definition used?	Did the instrument used have reliability and validity?	Was the same mode of collection used for all subjects?	Was the shortest length of prevalence period appropriate?	Were the numerators and denominators for the parameter of interest
First Author, Year						
Ballard, 2002	Yes	Yes	No	Yes	Yes	Yes
Cetinkaya, 2011	Yes	Yes	No	Yes	Yes	Yes
de Oliveira, 2019	Yes	No	No	Yes	Yes	Yes
Ferres-Amat, 2017	Yes	Yes	No	Yes	Yes	Yes
Flinck, 1994	Yes	Yes	No	Yes	Yes	Yes
Freudenberger, 2008	Yes	Yes	No	Yes	Yes	Yes
Friend, 1990	Yes	Yes	No	Yes	Yes	Yes
Harris, 1992	Yes	Yes	No	Yes	Yes	Yes
Hogan, 2005	Yes	No	No	Yes	Yes	Yes
Messner, 2000	Yes	Yes	No	Yes	Yes	Yes
Ngerncham, 2013	Yes	No	No	Yes	Yes	Yes
Perez-Aguirre, 2018	Yes	No	No	Yes	Yes	Yes
Puapornpong, 2014	Yes	Yes	No	Yes	Yes	Yes
Puapornpong, 2017	Yes	Yes	No	Yes	Yes	Yes
Ricke, 2005	Yes	Yes	No	Yes	Yes	Yes

First Author, Year	Overall Risk of Study Bias
Ballard, 2002	Moderate
Cetinkaya, 2011	Moderate
de Oliveira, 2019	High
Ferres-Amat, 2017	Moderate
Flinck, 1994	Moderate
Freudenberger, 2008	Moderate
Friend, 1990	Moderate
Harris, 1992	Moderate
Hogan, 2005	High
Messner, 2000	Moderate
Ngerncham, 2013	High
Perez-Aguirre, 2018	High
Puapornpong, 2014	Moderate
Puapornpong, 2017	Moderate
Ricke, 2005	Moderate



# Figure 1. PRISMA Flow Diagram

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

For more information, visit www.prisma-statement.org.

Figure 2. Overall Prevalence of Tongue-Tie (N = 24,536). Forest plot of the studies documenting prevalence of tongue-tie. The analysis included 15 studies with a total of 24,536 cases.



Figure 3. Prevalence of tongue-tie by diagnostic method (N = 24,536). Forest plot of studies comparing prevalence of tongue-tie by diagnostic method. The analysis included 15 studies with a total of 24,536 cases. Five studies (N = 10,110) used a formal diagnostic method; 10 studies (N = 14,426) diagnosed tongue-tie through visual examination.



Figure 4. A funnel plot of standard error by logit event rate.



Funnel Plot of Standard Error by Logit event rate

Figure 5. Prevalence estimates by year of publication. A figure demonstrating prevalence estimates of the 15 included studies by year of publication.



# References

# **Refer to Cumulative Reference List**

# Chapter III

Symptoms of problematic feeding in infants under 1 year of age undergoing frenotomy: A

review article

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This manuscript replaces the review of the literature section of the traditional dissertation. Rebecca Hill was the primary author on this paper and Dr. Pados was second author. Rebecca Hill completed the systematic review and synthesis under the supervision of Dr. Pados. The readership of this journal includes healthcare professionals, particularly providers such as physicians, nurse practitioners and physician assistants involved in the care of pediatric patients. This manuscript was published on July 12, 2020 in Acta Paediatrica, which has an impact factor of 2.265. This manuscript represents a significant contribution to the dissertation work.

#### Abstract

Aim: To identify and summarize original research addressing the impact of tongue-tie on infant feeding, comparing symptoms of problematic feeding before and after frenotomy. **Methods:** A literature search was conducted on frenotomy and infant feeding. Databases searched include CINAHL and PubMed. The search terms ((tongue-tie) or (ankyloglossia)) and ((feeding) or (breastfeeding) or (bottle-feeding)) and ((frenotomy) or (frenectomy) or (frenulectomy) or (frenulotomy)) were used. **Results:** Maternal nipple pain, breastfeeding self-efficacy, and LATCH scores improved after frenotomy. No complications of frenotomy were reported. Little data is available on the effect of frenotomy on the infant's feeding. The limitations of the current literature restrict our understanding of who benefits from frenotomy and which aspects of infant feeding improve after the procedure. Conclusion: Healthcare providers should add tongue-tie as a differential diagnosis when nipple pain is present despite position changes, latch adjustment, or lactation support. Providers should be trained in the appropriate method for screening and diagnosing tongue-tie. Issues with bottle-feeding, such as difficulty latching, excessive gas, or prolonged feedings may be related to tongue-tie. Improvement of infant outcomes after frenotomy would give healthcare providers evidence that frenotomy is a reasonable treatment for tongue-tie. Quantifiable data on physiologic changes resulting from frenotomy are needed.

Keywords: Ankyloglossia; Breast feeding; Bottle feeding; Feeding Behavior; Frenotomy

# Key Notes:

1. The current literature supports frenotomy to reduce short-term maternal nipple-pain

in breastfeeding mothers.

2. The evidence to date has not utilized comprehensive measures of infant feeding to evaluate the effects of frenotomy for the infant with tongue-tie.

3. Future research that focuses on physiologic changes in the infant post-frenotomy is needed to support the use of frenotomy for the treatment of tongue-tie.

## Introduction

Ankyloglossia, commonly referred to as tongue-tie, is a congenital anomaly where the tongue is abnormally tightly connected to the floor of the mouth, limiting its mobility (Messner, Lalakea, Aby, Macmahon, & Bair, 2000). This anomaly has been cited in literature dating back to the 1700s (Theobald, 1764) and was routinely treated up through the 1940's, when infant formula use increased and breastfeeding rates declined (Wright & Waterson, 2006). Prevalence of tongue-tie has been reported as low as 0.3 percent (Çetinkaya et al., 2011) to as high as 16 percent (Ngerncham et al., 2013), with a hereditary component and male predominance (Segal, Stephenson, Dawes, & Feldman, 2007).

Organizations such as the Centers for Disease Control and the World Health Organization provide recommendations for exclusive breastfeeding (EBF) for the first six months of life (World Health Organization, 2019). Healthy People 2020 sought to increase exclusive breastfeeding rates to 25.5 percent (Office of Disease Prevention and Health Promotion, 2019). As of 2016, the exclusive breastfeeding rate at six months in the United States was 25.4 percent (Centers for Disease Control and Prevention, 2018). Exclusive breastfeeding through six months remains an objective in the preliminary development of Healthy People 2030, signifying that additional work is needed to increase this rate for more mother-infant dyads to obtain the benefits associated with breastfeeding (Department of Health and Human Services, 2019). A greater understanding of why mothers stop breastfeeding sooner than planned may help increase breastfeeding rates.

There are many factors that lead to early cessation of EBF, with one of the most commonly reported reasons being nipple pain (Kent et al., 2015). Tongue-tie has been associated with nipple pain (Kotlow, 2004) and frenotomy to treat tongue-tie is thought to reduce breastfeeding issues. A previous analysis of existing literature in 2017 identified studies with small sample sizes, few RCTs, a lack of longitudinal data, and limited evidence on the effects on infant feeding, with the most notable improvement in maternal reports of nipple pain (O Shea et al., 2017). This previous review did not evaluate the quality of the measures used to evaluate infant feeding. Despite the low strength of the evidence, a recent study reported an 866 percent increase in frenotomy rates from 1997-2012 (Walsh, Links, Boss, & Tunkel, 2017). The impact of frenotomy on infant feeding difficulties needs to be studied (O Shea et al., 2017). The purpose of this systematic review was to identify and summarize original research addressing the impact of tongue-tie on infant feeding, comparing symptoms of problematic feeding before and after frenotomy. The quality of the feeding-related outcome measures and psychometric properties of the assessment tools used were also reviewed.

#### Methods

#### Search Method

In September 2019, a literature search was conducted to review the effects of frenotomy on infant feeding. Databases searched include the Cumulative Index of Nursing and Allied Health Literature (CINAHL) and PubMed. The search terms ((tongue-tie) or (ankyloglossia)) and ((feeding) or (breastfeeding) or (bottle-feeding)) and ((frenotomy) or (frenectomy) or (frenulectomy) or (frenulotomy)) were entered into each database. The search was limited to research on humans, published in the English language, full-text, and with children aged birth to 12 months. No limit was placed on the time of publication. Duplicate articles were removed.

# **Study Selection**

To be included, original research articles must have reported the effects of frenotomy

on feeding before and after the procedure. Articles were excluded if they were not original research, if child age at the time of treatment was over 12 months, or if data was not collected prior to and after frenotomy. Titles and abstracts were screened independently by both authors using the inclusion and exclusion criteria and discrepancies were resolved. Articles that appeared to meet inclusion and exclusion criteria or that could not be properly evaluated based on the abstract were reviewed in full text. Both authors then reviewed the full-texts articles for inclusion/exclusion independently and discrepancies were resolved.

## **Selection of Study Characteristics Extracted**

Studies that met criteria for inclusion were evaluated for individual study characteristics. Relevant study characteristics that were extracted included the study sample size and sample characteristics, ankyloglossia diagnostic assessment tools used, frenotomy method(s) used, feeding-related outcomes measured, time points for measurement, and overall findings. Note that there are multiple terms used to describe the revision of the frenulum in infants with tongue-tie (e.g., frenotomy, frenulotomy, frenectomy, and frenulectomy). In this manuscript, we use the term "frenotomy" to describe the revision of the frenulum.

### **Assessment of Psychometric Properties of Outcome Measures**

After the feeding-related outcome measures were identified from the studies that met inclusion criteria, a review of the literature was conducted for each of these assessment tools to identify the psychometric properties. A search was conducted in both PubMed and CINAHL using the full name of the assessment tool. The search was limited to humans, English-language, full-text, and with children aged birth to 12 months. Articles reporting on the psychometric properties of the feeding-related measures were used to evaluate the quality of assessments used for studying the effect of frenotomy on feeding to date.

#### Results

The selection process for the chosen literature is presented in the Preferred Reporting Items of Systematic Reviews and Meta-Analyses (PRISMA) flow diagram (Figure 1). Fiftyfive full-text articles were screened for eligibility. Of those, 20 articles met inclusion criteria. Table 1 highlights the main components of each included study.

## **Sample Size & Characteristics**

The median sample size of the studies was 58 (range 14 - 246). Four of the studies had 30 or fewer infants, while seven studies included more than 100 infants. Most of the studies recruited mother-baby dyads that presented with breastfeeding difficulty despite lactation support (Ballard, Auer, & Khoury, 2002; Braccio, Chadderton, Sherridan, & Upadhyaya, 2016; Buryk, Bloom, & Shope, 2011; Dollberg, Marom, & Botzer, 2014; Geddes et al., 2008; Ghaheri, Cole, Fausel, Chuop, & Mace, 2017; Ghaheri, Cole, & Mace, 2018; Illing, Minnee, Wheeler, & Illing, 2019; Khoo, Dabbas, Sudhakaran, Ade-Ajavi, & Patel, 2009; Miranda & Milroy, 2010; Muldoon, Gallagher, McGuinness, & Smith, 2017; Schlatter et al., 2019; Sethi, Smith, Kortequee, Ward, & Clarke, 2013; Srinivasan et al., 2019; Srinivasan, Dobrich, Mitnick, & Feldman, 2006; Todd, 2014; Wakhanrittee, Khorana, & Kiatipunsodsai, 2016), and four of the studies screened all infants born during the study period (Ballard et al., 2002; Hogan, Westcott, & Griffiths, 2005: Martinelli, Marchesan, Gusmao, Honorio, & Berretin-Felix, 2015; Schlatter et al., 2019). Not all studies listed explicit inclusion or exclusion criteria. Beyond breastfeeding difficulties, some studies restricted inclusion to certain infant age ranges, (Emond et al., 2014; Ghaheri et al., 2017, 2018; Illing et al., 2019; Khoo et al., 2009; Srinivasan et al., 2018, 2006) specific tongue-tie severity (Ballard et al., 2002; Buryk, Bloom, & Shope, 2011; Emond et al., 2014), or baseline LATCH score (Emond et al., 2014). Exclusion criteria included the presence of other oral anomalies (Dollberg et al., 2014; Emond

et al., 2014; Martinelli et al., 2015; Schlatter et al., 2019; Srinivasan et al., 2019), prematurity (Emond et al., 2014; Geddes et al., 2008; Ghaheri et al., 2017, 2018; Schlatter et al., 2019), medical comorbidities (Ghaheri et al., 2017, 2018b; Martinelli et al., 2015; Schlatter et al., 2019), maternal breast surgery (Ghaheri et al., 2017, 2018), and delivery complications (Martinelli et al., 2015).

There was variety in maternal and infant demographic information reported across studies. The most commonly reported information was infant age, sex, and birth weight. One study did not report infant age (Ballard et al., 2002), and sex was reported in 14 of the studies (Ballard et al., 2002; Buryk et al., 2011; Dollberg et al., 2014; Ghaheri et al., 2017, 2018; Monica Hogan et al., 2005; Illing et al., 2019; Khoo et al., 2009; Martinelli et al., 2015; Sethi et al., 2013; Sharma & Jayaraj, 2015; Srinivasan et al., 2019, 2006; Wakhanrittee et al., 2016). Birth weight was reported in four studies (Khoo et al., 2009; Miranda & Milroy, 2010; Srinivasan et al., 2019; Wakhanrittee et al., 2016), and current infant weight was reported in one (Wakhanrittee et al., 2016). Only three reported infant ethnicities. In all three, the majority of infants were white (Ghaheri et al., 2017, 2018; Illing et al., 2019). One study discussed how maternal ethnicity, education level and socioeconomic status differed between the overall population at the hospital and those that enrolled in the study, but did not report the actual data comparing the sample to the overall population (Emond et al., 2014). None of the other 19 studies reported maternal ethnicity, socioeconomic status, or education level. Maternal data collected included age (Illing et al., 2019; Srinivasan et al., 2018, 2006; Wakhanrittee et al., 2016), parity (Dollberg et al., 2014; Muldoon et al., 2017; Srinivasan et al., 2019), and medical history (Srinivasan et al., 2019, 2006). Family history of tongue-tie was reported in three studies (Khoo et al., 2009; Muldoon et al., 2017; Schlatter et al., 2019).

### Ankyloglossia Assessment Tools Used

Across the 20 studies, tongue-tie was diagnosed using differing methods. Four studies used the Hazelbaker Assessment Tool for Lingual Frenulum Function (Hazelbaker, 1993) (HATLFF) (Ballard et al., 2002; Buryk et al., 2011; Emond et al., 2014; Schlatter et al., 2019), one used Kotlow's diagnostic criteria (Kotlow, 1999; Illing et al., 2019), and one used the Coryllos Grading System (Coryllos, Genna & Salloum, 2004; Dollberg et al., 2014). Srinivasan used the Frenotomy Decision Rule for Breastfeeding Infants (FDRBI) in the 2006 publication (Srinivasan et al., 2006) and then used a revised version of this tool, called the Frenotomy Decision Tool for Breastfeeding Dyads (FDTBD), in their 2019 publication (Srinivasan et al., 2019). One study classified tongue-tie as mild, moderate, or severe based on their own definition without evidence for the definition (Wakhanrittee et al., 2016). Three of the studies used a combination of two different assessment tools (e.g., Kotlow's and Coryllos) (Ghaheri et al., 2017, 2018; Srinivasan et al., 2018). Nine of the studies did not report if a standardized diagnostic assessment measure was used (Braccio et al., 2016; Geddes et al., 2008; Hogan et al., 2005; Khoo et al., 2009; Martinelli et al., 2015; Miranda & Milroy, 2010; Muldoon et al., 2017; Sethi et al., 2013; Sharma & Jayaraj, 2015).

The diagnostic tools used assess different characteristics of tongue-tie. The HATLFF focuses on tongue movement, while the Coryllos Grading System classifies tongue-tie based on severity of restriction to the floor of the mouth. Kotlow's diagnostic criteria involves measurement of free tongue (portion not attached to the frenulum) in millimeters (Kotlow, 1999). The FDRBI evaluates both the mother and infant. The mother is assessed for pain/trauma with breastfeeding and infant is assessed for his/her ability to sustain a latch onto the breast and weight gain. The infant's tongue is examined for the presence of an anterior frenulum and tongue movement specific to elevation, cupping, and protrusion. There is no

score requirement for the FDRBI (Srinivasan et al., 2006). The newer version, the FDTBD, evaluates these same characteristics in mother and baby. In addition, feeding time and milk transfer is subjectively reported by the mother. Lateral tongue movement is assessed along with elevation, suction, and protrusion. The maternal and infant categories are scored and if one or more symptoms are present in both the mother and infant (score of two or more), frenotomy is recommended (Srinivasan et al., 2019).

To date, very little has been published on the psychometric properties of the assessment tools used to diagnose tongue-tie. The Kotlow, Coryllos, FDTBD, and FDRBI assessment tools have no published psychometrics. The HATLFF has demonstrated interrater reliability in the measurement of tongue mobility (elevation, extension, and lateralization) when comparing infants with and without tongue-tie (Amir, James, & Donath, 2006). However, researchers found that the HATLFF resulted in a large number of unclassified infants, potentially missing tongue- tie diagnosis (Madlon-Kay et al., 2008), as well as false negatives (Ricke et al., 2005). Others acknowledged subjectivity of the HATLFF between raters (Emond et al., 2014).

### **Frenotomy Methods**

Twelve of the studies included in this review used scissors as the method for frenotomy (Ballard et al., 2002; Geddes et al., 2008; Hogan et al., 2005; Illing et al., 2019; Khoo et al., 2009; Martinelli et al., 2015; Miranda & Milroy, 2010; Sethi et al., 2013; Sharma & Jayaraj, 2015; Srinivasan et al., 2006, 2019; Wakhanrittee et al., 2016), two used laser (Ghaheri et al., 2017, 2018), one reported "varied" methods (Muldoon et al., 2017), and five studies did not indicate the method of treatment (Braccio et al., 2016; Buryk et al., 2011; Dollberg et al., 2014; Emond et al., 2014; Schlatter et al., 2019). No complications of frenotomy were noted in any of the 20 studies.

## **Feeding-Related Outcomes**

The primary feeding-related outcomes measured included nipple pain, breastfeeding assessment, breastfeeding self-efficacy, and infant gastroesophageal reflux. Eleven of the 20 studies utilized subjective assessment of improvement in breastfeeding via maternal report (Ballard et al., 2002; Braccio et al., 2016; Dollberg et al., 2014; Hogan et al., 2005; Illing et al., 2019; Khoo et al., 2009; Martinelli et al., 2015; Schlatter et al., 2019; Sethi et al., 2013; Srinivasan et al., 2006, 2019). All eleven reported subjective improvements in breastfeeding, including decreased nipple pain, improvements in latch, and shorter feeding times. Subjective nipple pain improved immediately after frenotomy was performed. Dollberg stated 90% of breastfeeding problems were alleviated after frenotomy; however, three percent of women reported worsening of symptoms at two weeks post-frenotomy (Dollberg et al., 2014).

One study included breastfed and artificially fed infants. This study measured subjective outcomes on bottle feeding pre- and post-frenotomy, including total feeding time, amount of dribbling from the bottle nipple, and maternal report of infant gassiness (Hogan et al., 2005). Subjective improvements in feeding time, dribbling and gas were noted by the mothers of the eight infants in the treatment group with no improvements in the nine babies in the control group (p < .001). Wakhanrittee found that frenotomy performed 24 hours or less after birth (p < .001), less severe tongue-tie (p < .001), female sex (p = .01), and more children in the family (p = .02) significantly contributed to EBF success at three-months postfrenotomy (Wakhanrittee et al., 2016). Sharma and colleagues demonstrated similar findings, with mothers of infants less than 30 days old having greater improvements in breastfeeding symptoms, although the difference was not significant (p = .09) (Sharma & Jayaraj, 2015).

An increase number of sucks and a decrease in pause length between suck groups was noted in infants post-frenotomy when compared to infants that did not undergo the procedure. Geddes found that breastmilk production was significantly increased in the right breast, with changes in feeding mechanics on ultrasound after frenotomy. In this study, only six of the 24 mothers completed post-frenotomy milk transfer measurements. Pre-frenotomy feeding mechanics revealed biting on the nipple or an inability to maintain an effective seal, both of which cause nipple pain (Geddes et al., 2008).

Nipple pain. The most frequent outcome measured was nipple pain, evaluated in 19 of the 20 studies, and the one study that did not measure improvements in nipple pain mentioned it as the most common presenting symptom (Sharma & Jayaraj, 2015). Assessment of nipple pain was completed pre and post-frenotomy in all 19 studies, using a variety of assessment techniques. Pain rating using a visual analogue scale (VAS) (Ballard, Auer, & Khoury, 2002; Emond et al., 2014; Ghaheri, Cole, Fausel, Chuop, & Mace, 2017; Ghaheri, Cole, & Mace, 2018; Muldoon et al., 2017) or subjective maternal report (Braccio, Chadderton, Sherridan, & Upadhyaya, 2016; Dollberg et al., 2014; Hogan et al., 2005; Illing & Minnee, 2019; Khoo et al., 2009; Martinelli et al., 2015; Schlatter et al., 2019; Sethi, Smith, Kortequee, Ward, & Clarke, 2013) were the most commonly used methods to assess nipple pain. A Numeric Rating Scale (NRS) was used in four studies (Geddes et al., 2008; Miranda & Milroy, 2010; Srinivasan et al., 2019; Wakhanrittee et al., 2016), and the Short-Form McGill Pain Questionnaire (SF-MPQ) was used in two (Buryk et al., 2011; Srinivasan et al., 2006). In all 19 studies, there was a decrease in maternal nipple pain post-frenotomy, with improvements occurring immediately post-frenotomy and up through two-weeks post-procedure. The severity of tongue-tie was not correlated with the degree of pain or improvement in maternal

symptoms post-procedure (Hogan et al., 2005; Khoo et al., 2009). Khoo and colleagues (2009) found that mothers who presented with nipple pain as the main symptom of tongue-tie were more likely to be breastfeeding three months post- frenotomy (OR 5.8, 95% CI 1.1-31.6) (Khoo, Dabbas, Sudhakaran, Ade-Ajayi, & Patel, 2009).

**Breastfeeding Assessment.** In addition to the studies that reported subjective reports of breastfeeding, nine studies reported on breastfeeding assessment using a standardized measure. Three of the studies utilized the Infant Breastfeeding Assessment Tool (IBFAT) as a measure of frenotomy success (Buryk et al., 2011; Emond et al., 2014; Sharma & Jayaraj, 2015). Buryk, Bloom & Shope (2011) conducted a single-blinded RCT, measuring scores on the IBFAT immediately before and after frenotomy and again at two weeks post-procedure. Infants were randomly assigned to the frenotomy or control groups, with no statistically significant differences between groups prior to frenotomy (p = .44) (Buryk et al., 2011). The IBFAT scores improved significantly (p = .03) in the frenotomy group immediately after the procedure. Two- week comparison of IBFAT scores was not possible, as frenotomies were performed in all but one infant assigned to the control group. It is important to note that the version of the IBFAT used in this study consisted of four categories with a total possible score of 15, which is not consistent with the original tool development, that allows for a maximum score of 12 (Matthews, 1988).

Emond and colleagues (2014) conducted a randomized parallel group trial, evaluating breastfeeding at the time of tongue-tie diagnosis, five days after frenotomy and again at 8 weeks post-procedure (Emond et al., 2014). A control group was assigned, with 44 of 52 infants undergoing frenotomy before the 8-week follow-up (Emond et al., 2014). This RCT did not show any statistically significant improvements in breastfeeding via the IBFAT (p =

.76). Sharma & Jayaraj (2015) conducted IBFAT scoring via telephone survey pre-frenotomy and one-month post-frenotomy. A statistically significant improvement in breastfeeding was noted by mothers in the frenotomy group, compared to those whose infants did not have the frenotomy procedure (p < .001). The severity of tongue-tie was not consistently documented across studies, and long-term effects were not assessed. The study by Emond and colleagues evaluated tongue- tie severity using the HATLFF, however, only those with mild-moderate tongue-tie were included, limiting generalizability to that subset of the population (Emond et al., 2014).

Six studies utilized the LATCH tool (Emond et al., 2014; Geddes et al., 2008; Schlatter et al., 2019; Srinivasan et al., 2019, 2006; Wakhanrittee et al., 2016), with a seventh adopting a modified version of the LATCH that has not been validated (Muldoon et al., 2017). Srinivasan and colleagues assessed breastfeeding using the LATCH tool immediately before and after frenotomy, with a statistically significant improvement in LATCH scores following frenotomy (p < .001) (Srinivasan et al., 2006). There were no differences in LATCH scores in the Emond (2014) study between groups (Emond et al., 2014). Another study by Srinivasan and colleagues in 2019 assessed breastfeeding using the LATCH tool pre-frenotomy, immediately post- frenotomy and again at days 2, 7, and 14 post-frenotomy. This study demonstrated statistically significant increases in LATCH scores immediately post-frenotomy and again from days seven to 14 post-frenotomy (p < .001) (Srinivasan et al., 2019). Samples sizes were highly variable in these studies, ranging from 26 to 246, and lack longitudinal data beyond eight weeks post-revision. The studies by Srinivasan, Dobrich, Mitnick & Feldman (2006) and Geddes, et al. (2008) did not use a validated tool to diagnose tongue-tie, had a small sample sizes (N = 30 and 24, respectively), and lacked control groups (Geddes et al.,

2008; Srinivasan et al., 2006). As previously mentioned, while Emond (2014) was an RCT, the authors limited the sample to those with mild-moderate tongue-tie and infants less than two weeks of age. Muldoon and colleagues reported a significant improvement in LATCH scores post-frenotomy (MD -0.50, 95% CI -0.67 to -0.33), using a modified version of the tool. While breastfeeding improved and maternal pain decreased, the rate of formula use doubled at 1-month post-frenotomy (Muldoon et al., 2017).

Srinivasan (2019) utilized frenotomy alongside lactation counseling, with a small sample size (N = 30), with several participants lost to follow-up. It is not possible to determine the independent influences of frenotomy on outcomes as concurrent lactation support was also included. The study by Wakhanrittee and colleagues had the largest sample, consisting of 246 infants diagnosed with tongue-tie. LATCH scores were significantly increased at 24-hours and one-week post-frenotomy (p < .001). However, this study also lacked a control group and endorsed a possible selection bias of motivated mothers eager to breastfeed (Wakhanrittee, Khorana, & Kiatipunsodsai, 2016). Significant improvements in LATCH scores were also noted by Schlatter at 2.5 weeks post-frenotomy (p = .01). This same study also used the Bristol Breastfeeding Assessment Tool (BBAT), and demonstrated a significant improvement in the BF assessment post-frenotomy (p = .01), with results limited by a small sample size (N = 23) and loss of participants to follow-up (Schlatter et al., 2019).

**Breastfeeding Self-Efficacy.** The 14-item Breastfeeding Self-Efficacy short form (BSES-SF) was used in three of the 20 studies included in this systematic review (Emond et al., 2014; Ghaheri et al., 2017, 2018). Sample sizes in these three studies were higher, ranging from 54 (Ghaheri et al., 2018a) to 237 (Ghaheri et al., 2017). BSES-SF was administered pre-frenotomy in all three studies. Emond and colleagues (2014) administered the tool again at 5

days and 8 weeks post-procedure and found statistically significant improvements in scores from baseline to day 5 post-procedure (p = .002), with no differences between groups at the 8week measurement, noting that most infants in the control group had undergone frenotomy by this time point. Breastfeeding self-efficacy improved in mothers whose infants underwent frenotomy, with fewer infants being fed by bottle by the five-day follow-up compared with the control group. The two studies conducted by Ghaheri and colleagues (2017, 2018) evaluated BSES using the short-form immediately prior to frenotomy and again at one week and one-month post- procedure. Both studies demonstrated statistically significant improvements in scores at one week and one-month time points, compared with preprocedure scores (p < .001).

Infant Gastroesophageal Reflux. The two studies by Ghaheri and colleagues (Ghaheri et al., 2017, 2018) evaluated scores on the Infant Gastroesophageal Reflux Questionnaire – Revised (I-GERQ-R) pre-frenotomy and one week and one month post-frenotomy. Both studies revealed significant decreases in symptoms of gastroesophageal reflux at all time points after frenotomy (p < .001). Factors that may have led to improvement in GERD beyond frenotomy, such as dietary changes in the breastfeeding mother, use of acid-reduction medications, or maturation of the infant gastrointestinal tract were not described.

# Quality of the Feeding-Related Assessments Used

**Nipple Pain.** Standardized tools used to assess nipple pain included the VAS (Atiken, 1969), the NRS (Dworkin, Tuck & Farrar, 2005), and the SF-MPQ (Melzack, 2005). These three pain scales have been used in research for both acute and chronic pain, most commonly for the assessment of back pain, but have not been validated for use to measure maternal

nipple pain.

**Breastfeeding Assessment**. Three instruments were used to evaluate breastfeeding, the IBFAT (Matthews, 1988), the LATCH tool (Jensen, Wallace, & Kelsay, 1994), and the BBAT (Ingram, Johnson, Copeland, Churchill, & Taylor, 2015). The psychometric properties of these instruments were reviewed in the literature and have been summarized on Table 2. The IBFAT consists of four items: readiness to feed, rooting, fixing, and sucking (Matthews, 1988). It was developed to assess breastfeeding readiness and competence of infants and can be used by mothers and health care professionals. Scores range from 0 to 12, with 12 indicating the most effective feeding. Higher IBFAT scores have been associated with greater satisfaction with breastfeeding from the maternal perspective (Schlomer, Kemmerer, & Twiss, 1999).

The LATCH is a tool used to document breastfeeding assessment, focusing on the key areas of breastfeeding using the LATCH acronym: Latch, Audible swallowing, Type of nipple, Comfort (breast/nipple) and Hold (positioning) (Jensen et al., 1994). Scores range from 0 to 10, with scores of nine or ten representing a breastfeeding session that does not require intervention. The LATCH tool can be completed by mothers or health care providers assessing a breastfeeding session. Both the LATCH tool and the IBFAT have been criticized for their use in evaluating breastfeeding pre- and post-frenotomy, considered to not be specific enough to measure effects of tongue-tie or frenotomy on breastfeeding (Griffiths, 2004).

The BBAT was developed for use by breastfeeding support providers to evaluate breastfeeding proficiency specifically for mothers with tongue-tied infants (Ingram, Johnson, Copeland, Churchill, & Taylor, 2015). The BBAT is a 4-item tool measuring positioning, attachment to the breast, sucking and swallowing. Scores range from 0-8, with higher scores indicating greater proficiency with feeding. At the time the tool was developed, it was found to be correlated with BSES, and to have acceptable internal consistency and interrater reliability (Table 2) (Ingram, Johnson, Copeland, Churchill, & Taylor, 2015).

**Breastfeeding Self-efficacy**. Self-efficacy was originally defined by Bandura (1977) as "an individual's confidence in his or her perceived ability to perform a specific task or behavior" (Bandura, 1977). Dennis developed the self-efficacy framework specific to breastfeeding confidence (Dennis, 1999) and later, developed the Breastfeeding Self-Efficacy Scale (BSES) (Dennis & Faux, 1999) and then the short form (BSES-SF) (Dennis, 2003). Scores on the BSES- SF range from 0-70, with higher scores indicative of greater breastfeeding efficacy and confidence from the maternal perspective.

**Gastroesophageal Reflux.** The Infant Gastroesophageal Reflux Questionnaire (I-GERQ) (Orenstein, Cohn, Shalaby, & Kartan, 1993), and its revised form, the I-GERQ-R (Kleinman et al., 2006) measure symptoms of gastroesophageal reflux. The I-GERQ-R is a 12-item questionnaire, with item responses based on a 1-week recall. Higher scores indicate worse symptoms.

# Discussion

This systematic review identified 20 research studies that have evaluated the effect of frenotomy on feeding in infants with tongue-tie. The literature suggests that there are improvements in maternal symptoms such as nipple pain, LATCH scores and self-efficacy with breastfeeding after frenotomy, with greater improvements when frenotomy is performed before one month of age. Frenotomy is a minimally invasive procedure without major complications noted, and maternal improvements following the procedure were present in all of the studies reviewed. However, there are significant limitations to the current literature

that restrict our understanding of who benefits most from frenotomy, which methods of treatment should be used, and in what ways the infant may benefit from frenotomy with regards to feeding.

A major limitation of the current literature is that we do not have adequate data to know which mother-infant dyads benefit most from frenotomy. There are anatomical and functional variations across infants with tongue-tie, which likely impact whether or not feeding improves with frenotomy. The published tools to diagnose tongue-tie have not been validated for use and thus cannot be used by clinicians or researchers to consistently assess and grade tongue-tie. Without a good measure and without consistent measurement across studies, we do not know which type of tongue-tie anatomy and function are responsive to frenotomy intervention. Similarly, there is variation across breastfeeding mothers in terms of breast and nipple anatomy, which likely impact the infant's feeding mechanisms at breast. Studies that have assessed infant anatomy, but have not assessed maternal anatomy, have not fully evaluated the dyad if breastfeeding is the feeding outcome of interest. For bottle-fed infants, variation in the type of nipple and its flow rate may also impact an infant's ability to maintain an effective seal, ingest excess air, or adapt to the feeding device when the oral anomaly is present.

The studies included in this review did not consistently and comprehensively describe the study samples in a way to allow readers to understand to whom their results apply. It is unclear what subset of the population seeks treatment for tongue-tie, where referrals are generated, and if there are racial, ethnic, or socioeconomic differences within the population of infants with tongue-tie. Emond and colleagues noted a difference between the population of mother-baby dyads delivering at the hospital where data were collected and those who presented to the breastfeeding clinic but did not specifically describe these differences. National data in the United States has demonstrated that more diagnoses of tongue-tie and treatment with frenotomy occur in families with private insurance and in mid- to high-income areas (Walsh, Links, Boss, & Tunkel, 2017). Sociodemographic factors of the mothers and infants were not reported in any of the reviewed studies, a major limitation that fails to recognize societal differences that may be present in a country where universal health care and insurance coverage for frenotomy is not guaranteed.

The literature does not provide adequate data on the age of the infants at the time of frenotomy. The infant's age, and therefore the length of time that they have learned and practiced feeding with their natural anatomy, likely affects how they will feed post-frenotomy. There are many other factors, such as feeding method(s) prior to frenotomy, maternal milk supply, gestational age at birth, and mothers' prior experience with breastfeeding that also play a role in infant feeding and are important to know when evaluating to whom frenotomy provides benefit.

While experts in the field of tongue-tie recommend frenotomy via laser, research has not been conducted comparing the outcomes of treatment between laser and scissors. Two case reports have demonstrated advantages of using laser for frenotomy, including decreased bleeding, improved visibility, and greater ability to manipulate the tissue (Doshi, Shah, Khandge, & Sanghavi, 2010; Elanchezhiyan et al., 2013). It is reassuring that no complications were reported in any of the reviewed studies, with the majority using scissors to correct tongue-tie. Several studies did not report that exact method used for frenotomy, another limitation of the current research.

The literature provides information about only certain aspects by which infants and
mothers benefit from frenotomy with regards to feeding. Knowing that maternal nipple pain is one of the most frequent reasons for breastfeeding cessation, it is clear why this was a commonly reported outcome. Across studies, an improvement in nipple pain was reported. The findings by Khoo and colleagues suggest that resolution of nipple pain, as a direct complication of tongue- tie, has the greatest impact on long-term breastfeeding success (Khoo et al., 2009). However, the reduction of nipple pain cannot be solely attributed to frenotomy, as nipple pain may diminish over time without intervention or may improve with other interventions, such as involvement of a lactation consultant, treatment of thrush, or use of a nipple shield. Conversely, nipple pain that results from something unrelated to the infant's tongue-tie (e.g., untreated thrush) may not improve after frenotomy, even if tongue-tie was a contributing factor. Studies exploring the effects of frenotomy on tongue-tie need to document assessment of both the infant and maternal anatomy for other contributing factors by trained personnel and report concurrent treatment strategies that may impact the effect of frenotomy on infant feeding.

In addition to improvements in nipple pain, the reviewed studies found that breastfeeding self-efficacy improved with frenotomy. Breastfeeding self-efficacy is a known correlate to success with breastfeeding; however, it has little relevance on the effectiveness of frenotomy for tongue-tie. Confidence and self-efficacy with breastfeeding may help women overcome barriers with feeding but should not be an independent measure of frenotomy success.

The infant factors that were measured, specifically symptoms of GERD, suck quality, and milk transfer, improved. Suck quality and milk transfer were assessed in very small samples and further research on larger samples is needed. Aerophagia-induced gastroesophageal reflux has been associated with poor latching in the presence of tongue-tie (Siegel, 2017). These findings should be explored in larger samples, controlling for co-morbidities and variables that may contribute to the development of GERD.

The studies reviewed lack comprehensive evaluation of infant feeding. The infant feeding tools that were used contain very few questions. While this decreases the time it takes to evaluate feeding, they cannot comprehensively assess infant feeding characteristics or difficulties. The LATCH tool has poor predictive validity of breastfeeding problems (Schlomer, Kemmerer, & Twiss, 1999), differences in scoring between lactation consultants and mothers (Adams & Hewell, 1997), and three of the five assessments are directed at problems experienced by the mother, not the infant (type of nipple, comfort with feeding, and hold/infant positioning). The IBFAT has demonstrated poor predictive validity (Matthews, 1988; Schlomer, Kemmerer & Twiss, 1999). Despite the BBAT being designed to assess problems specifically related to tongue-tie, there has not been psychometric testing to fully evaluate this measure. It is difficult to determine when the greatest improvement is seen after frenotomy, with outcome measures being evaluated over varied time spans, both in-person and via telephone follow-up. Studies conducting follow-up assessments over weeks to months introduce recall bias.

## **Recommendations for Practice**

Despite the limitations of the studies reviewed, breastfeeding assessments and nipple pain improved post-frenotomy. The possibility of tongue-tie causing nipple pain and the evidence that frenotomy improves nipple pain should be considered by healthcare providers during feeding assessments and when mothers report prolonged pain with breastfeeding. It is reasonable for healthcare providers to add tongue-tie to the differential diagnosis when nipple pain is present and persists despite position changes, latch adjustment, and/or lactation support. Providers should be trained in the appropriate method for screening and diagnosing tongue-tie, with the understanding that an improved screening measure is necessary. Issues with bottle-feeding, such as difficulty latching, clicking, dribbling, excessive gas, or prolonged feedings also warrant oral assessment. Tongue-tie should be considered as a potential factor in the development of infant GERD. With the known adverse effects of acidreducing medications (Safe et al., 2016), an oral assessment to rule out tongue-tie as a potential cause of GERD is reasonable prior to initiation of these medications.

### **Recommendations for Research**

Future research must control for infant and maternal characteristics that may impact feeding, such as history of breastfeeding experience, infant prematurity, current infant age, and the presence of co-morbidities known to affect feeding (e.g., congenital heart disease, bronchopulmonary dysplasia). A valid, reliable, and standardized assessment tool must be developed to improve effectiveness of screening for the anomaly. Maternal report of nipple pain and breastfeeding difficulties are important; however, it is also necessary to conduct research that focuses on infant-specific feeding outcomes related to tongue-tie and frenotomy. This can be accomplished by using comprehensive feeding-related outcome measures that evaluate feeding difficulties. Use of a valid and reliable tool to assess problematic feeding would enhance the rigor of the research beyond measuring maternal symptoms or the subjective reporting of symptoms.

Information on improvement of infant outcomes specific to tongue-tie after frenotomy would give healthcare providers evidence that frenotomy is a reasonable treatment for tonguetie. Symptoms of problematic feeding, weight gain, suck quality and strength, and total amount of milk transferred per feeding would provide quantifiable data on physiologic changes resulting from frenotomy. Longitudinal data on the impact of tongue-tie on craniofacial formation, dental and orthodontic needs, and speech development are needed. Ideally, this research should be conducted on a sample size determined via power analysis with a matched control group. The ethical implications of delaying tongue-tie treatment must be recognized, as this has affected the ability to conduct rigorous randomized control trials in this population.

## Conclusion

The literature reviewed demonstrated effectiveness of frenotomy for tongue-tie from the maternal perspective using measures that do not comprehensively evaluate infant feeding. It is reasonable to add tongue-tie as a differential diagnosis when an infant presents with symptoms of GERD, excess gas, substantial dribbling from a bottle, or when a breastfeeding mother reports persistent nipple pain or difficulties with infant latch. The growing interest in this topic and anecdotal reports of improvements following frenotomy warrant research to examine the effectiveness of frenotomy for varied degrees of tongue-tie severity. Referral and treatment guidelines cannot be established without use of a valid and reliable diagnostic method and evaluation of infant feeding improvements post-frenotomy using a comprehensive feeding measure. The research on frenotomy demonstrates short-term improvements in maternal symptoms but does not provide strong evidence to support frenotomy as treatment for infant feeding difficulties.



# Table 1. Characteristics of Included Studies

Author/Year	Sample Characteristics	Assessment / Treatment	Outcomes Measured	Outcome Timepoints
Ballard, Auer, & Khoury (2002)	<i>N</i> = 123 93 male, 61 female	Hazelbaker / Scissors	Nipple Pain (VAS) Subjective Maternal Symptoms	Immediately pre and post- frenotomy
Braccio et al (2016)	N = 158 Median age at frenotomy 2 weeks (range 1 day-5 months)	Not stated / Not stated	Maternal report of feeding problems	Immediately pre-frenotomy, 48 hours post-frenotomy and additional follow-up around 4 weeks post-frenotomy
Buryk, Bloom & Shope (2011)	<ul> <li>N = 58</li> <li>Randomly assigned to frenotomy</li> <li>(30) or usual care (28), Mean age</li> <li>6 days (all ≤ 30 days old)</li> <li>38 males, 20 females</li> </ul>	Hazelbaker / Not stated	Nipple pain (SF-MPQ), IBFAT	Immediately pre and post- frenotomy and 2-week follow- up
Dollberg, Marom & Botzer (2014)	N = 244 Median age 14 days (range 1-135 days) 143 male, 101 female Nipple pain and latch difficulty led to referral	Coryllos / Not stated	Maternal report of feeding problems	Pre-frenotomy, 2 weeks, 3 months, and 6 months post- frenotomy
Emond et al (2014)	N = 107 Randomly assigned to frenotomy (55) or usual care (52) Mean age 62 days Excluded if Hazelbaker score < 6 (severe tongue-tie)	Hazelbaker / Not stated	LATCH, IBFAT, BSES-SF, Nipple pain (VAS)	Pre-frenotomy, 5-days and 8- weeks post-frenotomy
Geddes et al (2008)	N = 24 Mean age 33 days (+/- 28 days, range 4-131 days)	Not stated / Scissors	24-hour milk production, LATCH, nipple pain (VAS), tongue movement via ultrasound	Immediately pre-frenotomy and within 7 days post-frenotomy
Ghaheri et al (2017)	N = 237 Mean age 4.4 weeks 86% White 133 males, 104 females	Kotlow and Coryllos / Diode laser	BSES-SF, IGERQ-R, nipple pain (VAS)	Immediately pre-frenotomy, 1- week and 1-month post- frenotomy

Ghaheri, Cole & Mace (2018)	N = 54 Mean age 8.3 weeks (range 7 day – 37 weeks) 43 white 28 male, 26 female	Kotlow and Coryllos / Diode Laser	BSES-SF, IGERQ-R, nipple pain (VAS)	Immediately pre-frenotomy, 1- week and 1-month post- frenotomy
Hogan, Westcott & Griffiths (2005)	<ul> <li>N = 57, mean age 19 days (range 3-70)</li> <li>14 males, 14 females in treatment group</li> <li>1.3:1 male to female in control group</li> </ul>	Not stated / Scissors	Maternal report of feeding difficulties	Immediately pre-frenotomy and 24 hours, weekly x 4 weeks, and at 4 months Frenotomy group (n = 28) compared with control group (n = 29)
Illing et al (2019)	N = 176 Mean age 44 days 109 males, 67 females	Kotlow / Scissors	Maternal report of feeding problems	Immediately pre-frenotomy and approximately 3 weeks post- frenotomy (mean time 23 days)
Khoo et al (2009)	N = 62 40 males, 20 females Mean age 23.5 days (+/- 17.1)	Not stated / Scissors	Maternal reports of feeding symptoms and degree of difficulty	Immediately pre-frenotomy and 3-month follow-up
Martinelli et al (2015)	N = 28 20 male, 8 female Frenotomy at 45 days of age	Not stated / Scissors	Maternal report of feeding problems, number of sucks and length of pause between sucks	Day 30 of life (pre-frenotomy) and day 75 of life post- frenotomy, compared between frenotomy and control group (N = 14 each)
Miranda & Milroy (2010)	N = 51 completed study Age range 12-36 days	Not stated / Scissors	Infant weight, number of feeding sessions/24 hours, nipple pain (NRS)	Immediately pre-frenotomy and 2-week follow-up
Muldoon et al (2017)	N = 89 Mean age at frenotomy 7 weeks 3 days	Not stated / Varied	Nipple pain (VAS), modified LATCH	Immediately pre-frenotomy and 1-month post-frenotomy
Schlatter et al (2019)	N = 30 Characteristics of sample undergoing frenotomy not described	Hazelbaker / Not stated	LATCH, BBAT, maternal report of feeding problems	Immediately pre-frenotomy and 2.5 weeks post-frenotomy
Sethi et al (2013)	N = 52 Mean age 19 days (range 3-120) 35 male, 17 female All mothers had BF problems prior to referral	Not stated / Scissors	Maternal report of feeding symptoms, rate of EBF post- frenotomy	Pre-frenotomy and within 5 months post-frenotomy

Sharma & Jayaraj (2015)	N = 42 36 had frenotomy 23 males, 19 females Median age at treatment 38 days (range 15-178 days)	Not stated / Scissors	IBFAT	Immediately pre-frenotomy and 1-month post-frenotomy, compared with those who did not have frenotomy (n = 6)
Srinivasan, Dobrich, Mitnick,	N = 27 18 male, 9 female	Frenotomy Decision Rule for	LATCH, nipple pain (SF-MPQ), maternal feeding	Immediately pre and post- frenotomy and 3-month
& Feidman (2006)	Mean age 19 days (range 2-71)	Breastreeding Infants / Scissors	questionnaire	telephone survey
Srinivasan et al (2019)	N = 30 Mean age 37.9 days (range 9-80) 20 male, 10 female	Coryllos and Frenotomy Decision Tool for Breastfeeding Dyads / Scissors	LATCH, nipple pain (VAS), maternal report of improvement in BF	Immediately pre and post- frenotomy and 2, 7, and 14 days post-frenotomy
Wakhanrittee, Khorana & Kiatipunsodsai (2016)	N = 246 147 male, 99 female Median age at frenotomy 50 hours 142 had severe tongue-tie 180 had moderate tongue-tie	Mild; Moderate; Severe / Scissors	LATCH, nipple pain (NRS), infant feeding pattern, EBF rate at follow-up	Immediately pre-frenotomy, 24- hours, 1-week and 3-months post-frenotomy

Table 2.	Psychometric Propertie	es of Assessment Tools

Purpose and Intended User	Target Population	Items: Number and Constructs	Reliability	Validity
Rapid breastfeeding assessment	Healthy full-term infants	4 items: Positioning, Attachment, Sucking and Swallowing	Interrater ICC = .78 Cronbach's alpha =	Construct validity with breastfeeding self-efficacy scale (r
Provider or Parent	Tested in infants up to 10 weeks old		.67	= .57)
Assess infant competence with breastfeeding	Healthy, full-term infants	6 items: Signaling, Rooting, Fixing, and	77-91% interrater agreement	Does not support predictive validity or
Provider or Parent		Sucking Pattern	Reliability .2778	discriminate validity
				convergent validity (rs = .5663)
				Construct validity r = .69 with LATCH, .78- .86 with MBA
Breastfeeding assessment and determine areas for improvement/intervention	Has been used in full-term and preterm infants	4 items: Latch-on, Audible swallowing, Nipple comfort,	Interrater ICC = .78- .89 (Chapman)	Construct validity with IBFAT (r = .69- .71) and MBA (r =
Provider or Parent		Assistance with positioning	85-100% interrater agreement (Adams)	.6888) (Altuntas 2014 & Riordan 1997)
			Test-retest r = .78	2007
			(Riordan)	Poor predictive validity for maintaining BF after 6 weeks (rs < .26, Riordan 2001), and for maternal satisfaction and
	Purpose and Intended UserRapid breastfeeding assessmentProvider or ParentAssess infant competence with breastfeedingProvider or ParentBreastfeeding assessment and determine areas for improvement/interventionProvider or Parent	Purpose and Intended UserTarget Population UserRapid breastfeeding assessmentHealthy full-term infantsProvider or ParentTested in infants up to 10 weeks oldAssess infant competence with breastfeedingHealthy, full-term infantsProvider or ParentFerenter userBreastfeeding assessment and determine areas for improvement/interventionHas been used in full-term and preterm infantsProvider or ParentFerenter userBreastfeeding assessment and determine areas for improvement/interventionHas been used in full-term and preterm infantsProvider or ParentFerenter userProvider or ParentFerence userProvider or ParentFerence user <t< td=""><td>Purpose and Intended UserTarget PopulationItems: Number and ConstructsRapid breastfeeding assessmentHealthy full-term infants4 items: Positioning, Attachment, Sucking and SwallowingProvider or ParentTested in infants up to 10 weeks old6 items: Signaling, Rooting, Fixing, and Sucking PatternAssess infant competence with breastfeedingHealthy, full-term infants6 items: Signaling, Rooting, Fixing, and Sucking PatternProvider or ParentHas been used in full-term and preterm infants4 items: Latch-on, Audible swallowing, Nipple comfort, Assistance with positioningBreastfeeding or ParentHas been used in full-term and preterm infants4 items: Latch-on, Audible swallowing, Nipple comfort, Assistance with positioning</td><td>Purpose and Intended UserTarget PopulationItems: Number and ConstructsReliabilityRapid breastfeeding assessmentHealthy full-term infants4 items: Positioning, Attachment, Sucking and SwallowingInterrater ICC = .78 Cronbach's alpha = .67Provider or ParentTested in infants up to 10 weeks old6 items: Signaling, Rooting, Fixing, and Sucking Pattern77-91% interrater agreementProvider or ParentHealthy, full-term infants6 items: Signaling, Rooting, Fixing, and Sucking Pattern77-91% interrater agreementProvider or ParentHas been used in full-term and preterm infants4 items: Latch-on, Audible swallowing, Nipple comfort, Assistance with positioningInterrater ICC = .78- .89 (Chapman)Provider or ParentHas been used in full-term and preterm infants4 items: Latch-on, Audible swallowing, Nipple comfort, Assistance with positioningInterrater ICC = .78- .89 (Chapman)Provider or ParentHas been used in full-term and preterm infants4 items: Latch-on, Audible swallowing, Nipple comfort, Assistance with positioningInterrater ICC = .78- .89 (Chapman)Provider or ParentHas been used in full-term and preterm infants4 items: Latch-on, Audible swallowing, Nipple comfort, Assistance with positioning85-100% interrater agreement (Adams)Provider or ParentHas been used in full-term and preterm infants4 items: Latch-on, Audible swallowing, Nipple comfort, Assistance with positioning85-100% interrater agreement (Adams)</br></br></br></br></br></td></t<>	Purpose and Intended UserTarget PopulationItems: Number and ConstructsRapid breastfeeding assessmentHealthy full-term infants4 items: Positioning, Attachment, Sucking and SwallowingProvider or ParentTested in infants up to 10 weeks old6 items: Signaling, Rooting, Fixing, and Sucking PatternAssess infant competence with breastfeedingHealthy, full-term infants6 items: Signaling, Rooting, Fixing, and Sucking PatternProvider or ParentHas been used in full-term and preterm infants4 items: Latch-on, Audible swallowing, Nipple comfort, Assistance with positioningBreastfeeding or ParentHas been used in full-term and preterm infants4 items: Latch-on, Audible swallowing, Nipple comfort, Assistance with positioning	Purpose and Intended UserTarget PopulationItems: Number and ConstructsReliabilityRapid breastfeeding assessmentHealthy full-term infants4 items: Positioning, Attachment, Sucking 

					breastfeeding problems (rs = .06- .5, Altuntas 2014)
Breastfeeding Self- Efficacy-Short Form (Dennis, 2003)	Measurement of breastfeeding confidence Breastfeeding women	Healthy, full term infants	14 items: maternal confidence	Cronbach alpha = .89 (Wutke, 2007 - .97 (Dennis, 2003)	Predictive validity, with significant differences in score between breast and bottle-feeding women (p < .001, Dennis, 2003) Predictive validity, with higher scores postpartum correlated with EBF at 8 and 16 weeks postpartum (p = .003, Wutke, 2007) Construct validity with significant differences in scores between women with and without prior BF experience (p < .001, Dennis, 2003) and women with more children (rs = .19, p = .024, Oliver-Roig, 2012) Correlated with
					global self-efficacy index (r = .41, p <

					.001) and Stress Management Self- Efficacy Scale (r = .24, p = .005, Oliver- Roig, 2012)
I-GERQ-R (Kleinman 2006)	Evaluation of GERD symptom severity and treatment effectiveness	Healthy infants	12 items: frequency, amount and discomfort with spit	Cronbach alpha = .8687	Significantly difference scores between control
	Parent		up, refusal/stopping	ICC = .85 for test- retest reliability	and infants with GFRD ( $n < 01$ )
	lucit		crying/fussing,	recest rendshity	
			hiccups, arching	ICC = .63 for inter-	Correlated with
			back, stop breathing/color	rater reliability	caregiver daily symptom diary (p <
			changes		.001)

## References

## **Refer to Cumulative Reference List**

## Chapter IV

The influence of tongue-tie and frenotomy on maternal breastfeeding symptoms and problematic

infant feeding

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This manuscript replaces portions of the methods and results section of the traditional dissertation. Rebecca Hill will be the primary author on this paper; Dr. Karen Lyons is second author; Dr. Kelly-Weeder is third author; and Dr. Pados will be the senior author on this paper. This paper will be submitted to the Journal of Human Lactation, which is an indexed and peer-reviewed journal with an impact factor of 2.349. The readership for this journal includes physicians, nurse practitioners, nurses, and lactation professionals concerned with breastfeeding and lactation. This manuscript will be submitted following defense of the dissertation. This manuscript represents a significant contribution to the dissertation work.

## Abstract

**Background:** Available evidence on the utility of frenotomy to treat tongue-tie suggests that breastfeeding mothers benefit from correction in the short-term, with a notable reduction in nipple pain. However, there remains a major gap in the literature, as we do not have evidence to support tongue-tie correction to treat symptoms of problematic feeding in infants. No studies have comprehensively evaluated the effects of frenotomy on feeding difficulties in infants with tongue-tie using valid and reliable measures of infant feeding. Additionally, the relationship between maternal symptoms and symptoms of problematic infant feeding in the context of tongue-tie is unknown.

**Research aim/question(s):** In a sample of infants with tongue-tie undergoing frenotomy and their mothers, the aims of this study were to: 1) describe changes in symptoms of problematic feeding, as measured by the Neonatal Eating Assessment Tool (NeoEAT), pre- and post-frenotomy, 2) explore the contribution of infant age to the magnitude of change in problematic feeding pre- and post-frenotomy, 3) describe changes in maternal symptoms pre- and post-frenotomy, and 4) evaluate the relationships between maternal symptoms and symptoms of problematic feeding pre- and post-frenotomy.

**Methods:** Mother-infant dyads were recruited from a pediatric dental office in the northeast region of the United States between July and November 2020. Diagnosis of tongue-tie was made using Kotlow's diagnostic criteria. Mothers were asked to complete an online survey about their family history, infant's health and feeding, and maternal symptoms, if breastfeeding, on the day of the frenotomy and 2 weeks post-frenotomy. Subgroups of the sample were defined by mode of feeding (exclusive breastfeeding, exclusive bottle-feeding, or mixed feeding). Paired *t*-tests compared NeoEAT total and subscale scores between the two time points. Linear regression

evaluated the effect of infant age on NeoEAT scores. Logistic and Poisson regression were used to compare changes in maternal symptoms from pre- to post-frenotomy. Correlations between maternal symptoms and NeoEAT scores pre- and post-frenotomy were examined to identify which maternal symptoms were associated with NeoEAT scores as a priori criteria to include in the regression model. Symptoms that were identified as statistically significant were then evaluated using generalized linear modeling to explain the strength of the relationship between each symptom and NeoEAT scores. For analyses comparing maternal symptoms at both time points, only the subsample of mothers who were exclusively breastfeeding at both time points were included.

**Results:** A total of 102 participants completed the first survey and 84 completed the 2-week follow-up survey. NeoEAT scores were lower in all subgroups after frenotomy, with no association between infant age and changes in these scores. Both maternal and infant symptoms improved after tongue-tie correction. Maternal symptoms of painful and difficult latch, creased/cracked nipples, bleeding or abraded nipples, and chewing of the nipple were significantly less common after tongue-tie correction. Mothers who reported bleeding nipples, oversupply of breastmilk, and nipple thrush before tongue-tie correction demonstrated significantly higher NeoEAT subscale scores prior to frenotomy, suggesting infant symptoms of problematic feeding were associated with maternal symptoms.

**Conclusion(s):** In infants with severe tongue-tie who are less than 7 months old, this study found that both symptoms of problematic infant feeding and maternal symptoms decreased two weeks after frenotomy. These feeding symptoms improved in all modes of feeding used. Infant age was not a significant factor in whether infant feeding improved.

Keywords: ankyloglossia; breastfeeding; bottle feeding; frenotomy; feeding behavior

## Key Messages:

- With all three feeding methods (breast, bottle, mixed), there were significant reductions in NeoEAT total and subscale scores from pre- to post-frenotomy.
- Difficult or painful latching onto the breast, creased/cracked, bleeding, or abraded nipples, and gumming or chewing on the breast nipple, and incomplete breast drainage were significantly less common after tongue-tie correction.
- 3. Improvements in maternal and infant symptoms were present regardless of infant age at time of tongue-tie correction.
- 4. Using the validated NeoEAT instruments to evaluate feeding in conjunction with examination of both mother and infant will give providers additional data that may support specialty referral for tongue-tie evaluation and feeding support.

## Background

Ankyloglossia, also known as tongue-tie, occurs in approximately 8% of infants (Hill, Lee & Pados, 2020). The lingual frenulum is a normal anatomical structure, but tongue-tie occurs when the lingual frenulum develops abnormally, resulting in a tight connection between the tongue and base of the mouth, restricting tongue motion and function. The abnormal band of tissue pulls the tongue to the floor of the mouth (Messner, Lalakea, Aby, Macmahon, & Bair, 2000). This restriction impacts the ability of the tongue to produce the movement necessary to transfer milk from the breast or bottle to the mouth and into the posterior pharynx (Geddes et al., 2008) and prevents effective latch onto the breast (Kotlow, 2011). When latch is impaired, maternal nipple pain, decreased milk transfer, and infant feeding difficulties may ensue (Kent et al., 2015; Messner et al., 2020).

A recent consensus statement was published, acknowledging that tongue-tie may contribute to breastfeeding problems (Messner et al., 2020). Maxillary labial frenulum, also known as lip-tie, has been inconsistently defined. The impact of lip-tie on maternal symptoms or infant feeding problems has not been determined and its presence is considered a normal finding in infants (Messner et al., 2020). Buccal ties, defined as tight frenulum tissue inside the cheek, should not be corrected per the consensus statement (Messner et al., 2020). The consensus statement could not recommend a preferred grading system for tongue-tie severity due to the lack of validated assessment tools currently available (Messner et al., 2020). The most commonly used classification systems include the Kotlow Grading Scale (Kotlow, 1999), Coryllos Ankyloglossia Grading System (Coryllos, Genna & Salloum, 2004), and Hazelbaker's Assessment Tool for Lingual Frenulum Function (Hazelbaker, 1993). What constitutes mild to severe tongue-tie varies between assessment tools. Frenotomy is the gold standard treatment of tongue-tie, but the available evidence on the effects of frenotomy on the mother-infant dyad is lacking. In a recent systematic review, the evaluation of frenotomy in the treatment of tongue-tie has lacked the methodological rigor necessary to promote a change in screening, assessment, and treatment guidelines (O Shea et al., 2017). The main outcome variables that have been measured focus on maternal outcomes, namely nipple pain (Ghaheri et al., 2018b; Illing et al., 2019; Muldoon et al., 2017; Srinivasan et al., 2006). Analysis of current research suggests that frenotomy has greatest improvements in maternal symptoms when completed by one month of age (Hill & Pados, 2020), but we do not have comprehensive infant data to support this. Frenotomy results in short-term improvement in maternal nipple pain, but the effect of frenotomy on infant feeding has not been adequately studied using comprehensive, valid, and reliable measures (Hill & Pados, 2020).

The purpose of this study was to evaluate the effect of frenotomy on infant feeding and maternal symptoms. In a sample of infants less than 7 months old with tongue-tie undergoing frenotomy and their mothers, the aims of this study were to: 1) describe changes in symptoms of problematic feeding, as measured by the Neonatal Eating Assessment Tool (NeoEAT), pre- and post-frenotomy, 2) explore the contribution of infant age to the magnitude of change in problematic feeding pre- and post-frenotomy, 3) explore changes in maternal symptoms pre- and post-frenotomy, and 4) evaluate the relationships between maternal symptoms and symptoms of problematic feeding pre- and post-frenotomy.

#### Methods

This study was approved by the Institutional Review Board at Boston College. Design

This was a one-group pre/post intervention study design. We compared infant feeding

and maternal symptoms immediately before and two weeks after frenotomy.

## Setting

This study took place at a pediatric dentist office in northeast region of the United States between July and November 2020. There is one pediatric dentist at this location who conducts all of the assessments using the Kotlow criteria (Kotlow, 1999) and performs all of the frenotomy procedures, allowing for consistency of assessment and classification of tongue-tie.

## Sample

To be included in this study, the infant had to be less than 7 months old, diagnosed with tongue-tie, and undergoing frenotomy. Mothers needed to be proficient in English in order to be included and complete the surveys. We chose a cutoff age of less than 7 months because of the changes in feeding mechanics that occur with the introduction of solid foods around 6 months of age. Exclusion criteria were prematurity (born earlier than 37 weeks' gestation) (Zimmerman & Rosner, 2018), anomalies of the head, face, or neck that may impair feeding (e.g., cleft lip/palate) (Reid, 2006), or the presence of comorbid conditions associated with feeding difficulty (e.g., bronchopulmonary dysplasia) (Gewold, & Vice, 2006; Park, Knafl, Thoyre, & Brandon, 2015). Convenience sampling was used and all eligible mother-infant dyads during the study period were invited to participate.

## **Sample Size Calculation**

G\*Power 3.1.9.4 (G\*Power, Germany) was used to conduct an a priori power analysis. Independent t-test was used to calculate the required sample to achieve 90% power, given alpha of .05 (two-tail), using a standardized effect size method (Cohen's d) to detect a medium change in differences between means (0.5). The power analysis determined that a sample size of 84 would be sufficient. To obtain a final sample of 84 with both pre- and post-frenotomy data, using a conservative 40% response rate for post-frenotomy, we aimed to recruit 125 mother-infant dyads. Recruitment ended when 84 participants were recruited that had completed both pre- and post-frenotomy surveys.

## **Frenotomy Intervention**

Participation in this research study did not alter the course of clinical care provided to infants. Per standard of care at this dental practice, the dentist obtained informed consent for the frenotomy prior to the procedure. The infant was moved to a laser-safe patient room. Protective eyewear was applied to the infant who was then swaddled for the procedure and an office staff member (dental hygienist) held the infant's head to prevent movement during the procedure. Frenotomy was preformed using a 1,064-nm InGaAsP semiconductor diode laser. The tongue was elevated with a grooved director and the laser tip applied to the frenulum. A diamond-shaped wound after incision was considered a full frenulum release. The baby was then brought back to the mother and offered feeding via breast or bottle. Post-procedural stretching exercises were taught to the mother, to be completed 4 times per day for 4 weeks.

#### **Data Collection**

Study data were collected and managed using REDCap electronic data capture tools hosted at Boston College (Harris, Taylor, Thielke, Payne, Gonzales, & Conde, 2009; Harris, Taylor, Minor, Elliott, Fernandez, et al, 2019). REDCap (Research Electronic Data Capture) is a secure, web-based software platform designed to support data capture for research studies, providing 1) an intuitive interface for validated data capture; 2) audit trails for tracking data manipulation and export procedures; 3) automatic export procedures for seamless data downloads to common statistical packages; and 4) procedures for data integration and interoperability with external sources. Tongue-tie classification and maternal symptoms pre-frenotomy were extracted from the medical record by the first author after the mother agreed to participate in the study. The mother completed the pre-frenotomy survey on the day of the frenotomy procedure on a tablet provided by the research team. Baseline maternal information collected included maternal age, race/ethnicity, marital status, pregnancy and birth history, and prior experience with breastfeeding. Baseline infant data included gestational age at birth, current age at the time of frenotomy, sex, current feeding mode and medical status, inquiring specifically about diagnoses such as gastroesophageal reflux, endocrine, cardiac, or respiratory disorders. Two weeks post-frenotomy, mothers received an electronic survey via email which asked about changes in infant medical history and maternal symptoms since treatment of their infant's tongue-tie. At each time point, mothers completed the NeoEAT version (Breastfeeding, Bottle-feeding, or Mixed Feeding) that reflected the mode(s) of feeding used in the prior week.

## Measures

**Tongue-Tie Classification**. In this study, Kotlow's diagnostic criteria was used to gauge the severity of tongue-tie. This tool measures the length of the freely-mobile tongue in millimeters (Table 1), classifying tongue-tie severity from least restrictive (Class 1, mild, >16 mm free tongue) to most restrictive (Class IV, complete, < 3mm free tongue) (Kotlow, 1999). At the time of manuscript production, the psychometric properties of this assessment tool had not yet been studied.

**Infant Feeding**. Symptoms of problematic feeding were measured using the Neonatal Eating Assessment Tool (NeoEAT), a parent-report measure of feeding intended for use with infants under 7 months old. There are three versions of the NeoEAT: the NeoEAT – Breastfeeding, for infants who have exclusively breastfed in the past week; the NeoEAT –

Bottle-feeding, for infants who have exclusively bottle-fed in the past week; and the NeoEAT – Mixed Feeding, for infants who have both breastfed and bottle-fed in the past week. Each question on the NeoEAT is rated on a 5-point Likert scale, with answer options ranging from "never" to "always". Scores are summed to create a total score as well as subscale scores, with higher scores indicating more symptoms of problematic feeding. Descriptors for each subscale are summarized on Table 2. All three tools have been validated for and have demonstrated construct validity with the Infant Gastroesophageal Reflux Questionnaire and Infant Gastrointestinal Symptoms Questionnaire (Park, Thoyre, Pados, & Gregas, 2019; Pados, Thoyre & Galer, 2019).

The NeoEAT – Breastfeeding has 62 items and has excellent internal consistency reliability ( $\alpha = .92$ ) and test-retest reliability (r = .91) (Pados, Thoyre, Estrem, Park, & McCormish, 2018). The NeoEAT – Bottle-feeding consists of 64 items, with five subscales. This version also has high internal consistency reliability ( $\alpha = .92$ ) and test-retest reliability (r = .90; p< .001) (Pados, Thoyre, Estrem, Park, & McComish, 2018). The NeoEAT – Mixed Feeding is a 68-item scale with five subscales that has demonstrated strong reliability ( $\alpha = .88$ ) and test-retest reliability (r = 0.91, p< .001) (Pados, Thoyre & Galer, 2019). Norm-reference values have been developed (Pados, Park, & Thoyre, 2019a, 2019b).

**Maternal Symptoms**. If breastfeeding, mothers were asked about maternal symptoms at both time points. A checklist was adopted from the medical record form used in the dental office. Mothers indicated if they experienced any of the following symptoms: creased, cracked or blanching of their nipples; painful latch onto the breast; gumming or chewing of their nipples; infant unable to achieve a tight latch; poor or incomplete breast drainage; infected or abraded nipples; plugged ducts; mastitis; nipple thrush; over or under supply; or feelings of depression. A total sum of all maternal symptoms was calculated both pre- and post-frenotomy.

**Frenotomy Experience.** Participants were asked if the infant experienced any complications after the frenotomy, and if they would make the same decision to have the tongue-tie corrected based on their experience by the 2-week follow-up.

## **Data Analysis**

All data were analyzed in SPSS v.25 (IBM, 2017). Descriptive statistics were used to characterize the study population. Reliability analyses of each NeoEAT scale at both time points were evaluated using Cronbach's alpha. Figure 1 demonstrates which subsamples were used in the analyses for each aim. For Aims 1 and 2, data from mother-infant dyads who completed both time points were included. Mean scores for NeoEAT total and subscale scores were calculated for participants who reported the same mode of feeding at both time points. For Aims 3 and 4, only data from breastfeeding dyads were included because these aims involved maternal symptoms, which were only relevant if the mother was breastfeeding. Aim 3 included mothers who reported breastfeeding in any capacity at time one or time two, while Aim 4 included mothers that were exclusively breastfeeding at either time point.

For Aim 1, we used paired t-tests pre- and post-frenotomy for the NeoEAT total score (across all infants) and sub-analyses by feeding method (i.e., breast, bottle or mixed) for the subscale scores. Tests of normality were run for NeoEAT total and subscale scores. For scores that were not normally distributed, nonparametric tests (Wilcoxon Signed-Rank Test) were used to compare scores between the two time points. For Aim 2, a change score was calculated for the NeoEAT score by subtracting pre- from post scores. We used linear regression to determine the effect of infant age (in weeks) on change score for NeoEAT total scores and for each of the subscale scores.

In Aim 3, logistic regression was used to explore maternal symptoms pre- and postfrenotomy for each individual symptom. We used Poisson regression to determine if there were significant changes in the number of symptoms pre- to post-frenotomy. For aim 4, the relationship between maternal and infant symptoms were compared for exclusively breastfeeding mothers. This analysis included any participant who reported exclusive breastfeeding either preor post-frenotomy. Correlations were performed for each maternal symptom and NeoEAT total and subscale scores to examine which maternal and infant symptoms were related at each time point. The strength of the relationship between maternal symptoms and NeoEAT scores was evaluated using general linear modeling, excluding variables that were highly correlated with each other (p < .05). Statistical significance was set at  $\alpha = .05$  (two-tailed) for all statistical tests.

#### Results

A total of 102 mothers completed the pre-frenotomy survey and 84 completed the postfrenotomy survey for a study completion rate of 82.4%. There were no differences between respondents and non-respondents that could explain results of the follow-up survey (e.g., race/ethnicity, mode of feeding, tongue-tie severity). Participants were from five northeast states (NY, PA, VT, CT, NH), with parent age ranging from 19-45 years (M = 31.6, SD = 5.2). Most were married, in 2-parent households. The majority of participants were white, with a variety of educational backgrounds and income levels. Infant age ranged from 3 days to 29 weeks, 3 days (M = 6 weeks, SD = 6.23). The majority of infants (n = 92, 90.2%) presented with a Class III tongue-tie, indicating "severe" restriction with 3-7mm of free tongue based on Kotlow's criteria. Table 3 describes the full sample. Tables 4 describes the subsamples of breastfeeding, bottlefeeding and mixed feeding participants.

From pre-frenotomy to post-frenotomy, 24 mothers (25%) changed the method of feeding

their infant. Of this subsample, 17 (70.8%) increased breastfeeding. Fourteen of the 24 mothers (58.3%) switched from mixed breast/bottle feeding to exclusive breastfeeding, seven (29.2%) changed from breastfeeding to mixed feeding, two (8.3%) changed from bottle to breastfeeding and one (4.2%) switched from bottle to mixed feeding.

While the anomaly of interest for this dissertation was tongue-tie, all infants in this study were also diagnosed and treated for lip tie. Approximately one-third (35%) of infants were also diagnosed with buccal ties, although none of these were corrected. Of the 84 respondents who completed the post-frenotomy survey, 82 answered questions about recovery and decision to treat their infant's tongue-tie. One respondent questioned the possibility of "reattachment" of the frenulum, while 81 indicated no complications. Almost all (n = 79 of 82, 96.3%) would make the same decision again to have the frenotomy performed, while one responded they would not treat again, and two were unsure if they would make the same decision to treat. These respondents did not provide additional information about why they would not treat again.

#### **Psychometric Properties of NeoEAT**

Reliability of the NeoEAT scales were assessed both pre- and post-frenotomy (Breastfeeding, Bottle-Feeding, and Mixed Feeding NeoEAT instruments). Internal reliability was excellent for all scales. Cronbach's alpha for the NeoEAT Breastfeeding scale was  $\alpha = .94$ pre-frenotomy and  $\alpha = .90$  post-frenotomy. Reliability for the Bottle-Feeding scale was  $\alpha = .98$ pre-frenotomy and  $\alpha = .94$  post-frenotomy. The Mixed Feeding NeoEAT reliability was  $\alpha = .96$ and  $\alpha = .94$  for pre- and post-frenotomy, respectively.

## Aim 1

Differences in scores by group are provided on Table 5. In the group of infants who were exclusively breastfed at both time points (n = 23), NeoEAT total score decreased by 27.8 points

from pre- to post-frenotomy (p = .003, d = .7). Subscale scores decreased significantly from preto post-frenotomy for the Infant Regulation (p = .002, d = .7), Energy and Physiologic Stability (p = .001, d = .8), Oro-Pharyngo-Esophageal (p = .008, d = .6), and Feeding Efficiency and Sensory Responsiveness (p = .008, d = .6) subscales.

In the bottle-feeding only group (n = 10), NeoEAT – Bottle-Feeding total score decreased from pre- to post-frenotomy by 44.6 points (p = .03, d = 1). There were significant decreases in the Energy and Physiologic Stability (p = .002, d = 1.4), Gastrointestinal Function (p = .02, d = .9), and Sensory Responsiveness (p = .03, d = .9) subscales.

In infants who were feeding by both breastfeeding and bottle-feeding (n = 27), the NeoEAT – Mixed Feeding total score decreased by 31.1 points (p = .001, d = .7). The Infant Regulation (p = .002, d = .7), Energy and Physiologic Stability (p = .03, d = .5), Gastrointestinal Function (p = .001, d = .7), and Feeding Flexibility (p = .002, d = .7) subscales all decreased from pre- to post-frenotomy.

## Aim 2

In the breastfeeding-only (n = 23) and bottle-feeding-only groups (n = 10), there was no association between infant age and change scores of the NeoEAT total or subscale scores. For infants fed by both breast and bottle (Mixed, n = 27), the implementation of new reflux strategies (e.g., elevating head of bed, holding upright after feeding) was associated with higher infant regulation subscale scores ( $\beta = 6.66$ , p = .005). Taking into account this significant confounding variable, older age at time of frenotomy was also significantly associated with higher infant regulation scores post-frenotomy ( $\beta = .353$ , p = .01). Table 6 presents results of analyses for aim 2 in each subgroup.

Mothers who were breastfeeding in any capacity (n = 85) reported up to 8 concurrent symptoms pre-frenotomy, with the most common symptoms being painful latching (n = 61), difficulty achieving successful latch (n = 50), and gumming/chewing of the nipple while eating (n = 49). All reported maternal symptoms decreased from pre- to post-frenotomy. Postfrenotomy, 75 mothers were breastfeeding. The most common symptoms were painful latch (n =21), gumming/chewing of the nipple (n = 17), and incomplete breast drainage (n = 14). Using Poisson loglinear modeling, the likelihood ratio chi-square test indicated that the full model, which included total maternal symptom count, was a significant improvement in fit over a null (no predictor) model (p < .001). The number of maternal symptoms was significantly different pre- to post-frenotomy ( $\beta = .881$ , SE = .1062, p < .001). The predicted number of maternal symptoms was .881 greater pre-frenotomy. The incidence rate ratio (ExpB) indicates that for every one-unit decrease on the predictor (time), the incidence rate of maternal symptoms increased by a factor of 2.414. In other words, the incidence rate of maternal symptoms was 141% higher before frenotomy ((2.414-1) \*100). Comparison of symptoms at both time points are presented on Table 7.

Seven of the 15 symptoms assessed had significantly greater likelihood of occurring prior to frenotomy. Poor infant latch or abraded nipples were eight times more likely prior to frenotomy (OR = 8.312, p < .001, OR = 8.763, p = .04). Odds of having painful latching onto the breast was seven times greater prior to frenotomy (OR = 6.536, p < .001). Gumming or chewing on the breast nipple or bleeding nipples were five times more likely to occur before treatment of infant tongue tie (OR = 4.644, p < .001 and OR = 4.792, p = 001, respectively). Table 8 explains the findings for each maternal symptom.

Aim 4

There were 35 mothers who reported exclusive breastfeeding pre-frenotomy and 39 who were exclusively breastfeeding post-frenotomy. Prior to frenotomy, three maternal symptoms correlated with NeoEAT scores. After frenotomy, some of the maternal symptoms that persisted correlated with NeoEAT – Breastfeeding total and subscale scores. Tables 9 and 10 depict pre-and post-frenotomy symptoms that correlated with NeoEAT scores.

Creased, cracked or blanched nipples, bleeding nipples, and gumming/chewing of the nipple were excluded for multicollinearity with painful latching onto the breast. Before tonguetie correction, oversupply was significantly associated with increased total NeoEAT score (28.9point increase, t = 2.503, p = .017), Energy and Physiologic Stability (6.4 points higher, t = 2.368, p = .024), Feeding Efficiency and Sensory Responsiveness (4.5 points higher, t = 2.656, p = .012), Oro-Pharyngo-Esophageal (7.5-point increase, t = 2.408, p = .022), and Compelling Symptom (2.5 points higher, t = 2.452, p = .020) subscale scores. Feeding Efficiency and Sensory Responsiveness subscale score increased by 4.9 points (t = 2.730, p = .010) when mothers reported poor latch onto the breast.

Following frenotomy, the total NeoEAT score was 35.4 points higher (t = 2.393, p = .027) in mothers reporting painful latching and 24.5 points higher (t = 2.419, p = .025) in mothers reporting incomplete breast drainage. Infant Regulation subscale scores were 15.1 points higher in mothers with painful latch (t = 4.436, p < .000). Maternal report of incomplete breast drainage was significantly associated with higher Energy and Physiologic Stability scores (7.9 points higher, t = 3.523, p = .002), Oro-Pharyngo-Esophageal scores (6.2 points higher, t = 2.142, p = .044), and Compelling Symptoms of Problematic Feeding (2.4 points higher, t = 2.644, p = .015). There were no maternal symptoms associated with changes in Gastroesophageal Function, Gastrointestinal Function, or Feeding Efficiency and Sensory

Responsiveness subscale scores.

## Discussion

This study is the first to use a valid and reliable measure of infant feeding to evaluate change in symptoms of problematic feeding after frenotomy in infants with tongue-tie. It is also the first to explore the relationship between infant feeding and maternal symptoms in breastfeeding mothers in the context of frenotomy for tongue tie.

It is important to acknowledge the transition to exclusive breastfeeding after frenotomy in 16 mothers (19% of the final sample) who were using bottle-feeding or a mixed approach to feeding before tongue-tie correction. There was also an increase in breastfeeding in those who were bottle-feeding prior to frenotomy, with 12% switching to exclusive breastfeeding or mixed feeding. There were no complications associated with the procedure, although one participant mentioned the possibility of regrowth of the frenulum tissue. Nearly all participants would elect to have the procedure again. This is important information for health care providers; mothers who present with feeding difficulties perceive improvements related to correction of severe tongue-tie. Respondents who completed the follow-up survey were less likely to have prior experience with breastfeeding (t = 2.063, p = .04) compared to non-respondents.

In all three feeding methods, there were significant reductions in NeoEAT total and subscale scores pre- to post-frenotomy regardless of infant age at the time of correction. These results suggest that, regardless of age, improvements are seen in symptoms of problematic feeding after frenotomy in infants with severe tongue tie. The sample sizes of infants in the 2 - < 4-month (n = 12) and 4 - < 7-month (n = 8) age groups were small. Future research should aim to increase the variation in infant age and sample sizes within these subsamples to further evaluate differences in feeding and changes in feeding symptoms after frenotomy.

With re-evaluation two weeks following the procedure, and no changes to medical history or infant medications during this time, it is likely that the improvements in symptoms of problematic feeding can be attributed to correction of the infant's tongue tie. Understanding how tongue-tie and its correction may lead to improvements in an infant's ability to self-regulate. become more efficient with eating, and reduce gastrointestinal discomfort provides critical information for parents and providers when faced with problematic feeding symptoms in infants. Differences in effect sizes between the three modes of feeding suggest that greatest improvements in symptoms of problematic feeding occurred in the bottle-fed subsample. The sample size of bottle-fed infants was small, so further exploration of this phenomenon in future studies is warranted. The infants with higher Infant Regulation scores post-frenotomy likely required additional strategies to manage these symptoms beyond tongue-tie correction. Using the validated NeoEAT instruments to evaluate feeding alongside physical examination will give providers additional data that may support specialty referral for tongue-tie and feeding support. As this is the first study to comprehensively evaluate symptoms of problematic feeding in infants with severe tongue-tie, future research would be strengthened by continuing to evaluate infants with a valid and comprehensive measure to better understand how tongue-tie impairs feeding.

Similar to other studies, maternal symptoms significantly decreased post-frenotomy. The most common symptoms were consistent with those described in previously published research (Hill & Pados, 2020). For mothers reporting these symptoms, evaluation of a breastfeeding session to examine latch, position, and infant ability to sustain a breastfeeding session will assist providers in identifying differential diagnoses such as tongue-tie, or breastfeeding mechanics that can be adjusted to reduce maternal discomfort. With the significant decrease in maternal symptoms post-frenotomy without other intervention, tongue-tie should be considered as a

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potential cause of painful or difficult latching or infant chewing on the nipple during feeding. An interdisciplinary evaluation with providers and lactation counselors would be beneficial to address positioning or other factors that may cause these symptoms.

The association between maternal symptoms and infant feeding challenges helps explain how the maternal experience can reflect infant feeding problems and vice versa. Conceptually, it is reasonable to expect that oversupply of breastmilk would impact Energy and Physiologic Stability, Oro-Pharyngo-Esophageal Function, Feeding Efficiency and Sensory Responsiveness, and Compelling Symptoms of Problematic Feeding. These NeoEAT – Breastfeeding subscales are related to how an infant responds to breastfeeding. An infant needing to take frequent breaks while eating, being unable to handle how fast milk comes out of the breast, drooling breastmilk from the sides of the mouth are findings consistent with oversupply (Fisher, n.d.). Impaired tongue mobility due to tongue-tie may be a reason for mothers to feel fullness/excess breastmilk and concurrent inability of an infant to adapt to large supply. Gagging on the breast or frequent feeding can be related to ineffective latching, as both symptoms may reflect an infant's inability to sustain a breastfeeding session (Garbarino et al., 2013).

While many mothers reported improvement in symptoms, some persisted following frenotomy to treat severe tongue-tie. In those that continued to report painful latch onto the breast or incomplete breast drainage, post-frenotomy NeoEAT scores were higher. This may suggest that the infant needs additional time to re-learn proper breastfeeding or that other factors are present beyond tongue-tie causing these persistent maternal symptoms. Evaluating maternal symptoms and infant feeding longitudinally, in conjunction with feeding support mothers receive would provide additional information on why and how tongue-tie correction or other factors may reduce these symptoms over time. This information adds to our current knowledge about severe tongue-tie and how this anomaly affects both the mother and the infant. In the research published to date, the outcomes of interest have been either the mother or the infant but lack dyadic evaluation necessary for the feeding pair. A better understanding of how the dyad is affected by severe tongue-tie gives insight to providers of symptoms the mother or the infant may have if severe tongue-tie is present and how the combination of these symptoms may result in early breastfeeding cessation or continued feeding challenges.

## Limitations

This study should be viewed in light of several limitations. First, the sample was homogenous, representing mainly white mother-baby pairs in the northeast region of the United States. We do not have data to compare respondents to non-respondents and do not know if the follow-up survey results would have been significantly different in the 18 participants who did not complete the follow-up survey. We also do not have information on how many mother-baby dyads presented to the office between July and November who did not have a diagnosis of tongue-tie, who declined treatment of the anomaly, or who declined to participate in this study.

With the majority of the sample diagnosed with Class III tongue-tie, we do not have information on how other degrees of tongue-tie severity might alter maternal and/or infant symptoms associated with the condition. All infants who were treated and included in this study were also diagnosed and treated for lip tie, so we cannot determine if it was the tongue-tie, liptie, or a combination of both oral restrictions that contributed most to the improvements in maternal and infant symptoms post-frenotomy. Future research should be directed at a more diverse sample, with a variety of tongue-tie severity classifications, and specific evaluation between infants with and without concomitant lip-tie. The small sample size of the bottle-feeding group is an important consideration when exploring changes to problematic feeding symptoms in these infants. Longitudinal evaluation of maternal and infant symptoms that includes a control group of infants without tongue-tie would strengthen our understanding of how this oral anomaly impacts feeding. Despite these limitations and the exploratory nature of the study, these findings make an important contribution to our understanding of the impact of frenotomy on the mother-infant dyad.

## Conclusion

In infants with severe tongue tie, this study found that frenotomy resulted in significantly decreased symptoms of problematic feeding in infants across all feeding methods, as well as significantly decreased maternal symptoms in breastfeeding mothers. Two-weeks after frenotomy, 70.8% of the sample reported having increased the amount of breastfeeding that they were doing; some mothers were able to transition from mixed breastfeeding and bottle-feeding to exclusive breastfeeding after frenotomy. In breastfeeding mother-infant dyads, symptoms of problematic feeding and maternal symptoms are highly inter-related. Significantly higher NeoEAT scores were present when mothers reported more severe maternal symptoms such as bleeding nipples or painful latch. Comprehensive evaluation of the breastfeeding mother-infant dyad needs to include assessment of both the mother and the infant, using valid and reliable measures whenever possible. Similarly, future research evaluating the efficacy of frenotomy in breastfeeding mother-infant dyads must acknowledge that the mother and baby are inextricably linked partners in feeding and must comprehensively evaluate the dyad. In infants and/or mothers with persistent symptoms two weeks after frenotomy, further evaluation and intervention is warranted.





Note. EBF = Exclusive breastfeeding.

Category	Length of the free tongue (mm)
Normal range of free tongue	>16
Class I: Mild ankyloglossia	12-16
Class II: Moderate ankyloglossia	8-11
Class III: Severe ankyloglossia	3-7
Class IV: Complete ankyloglossia	<3

Table 1. Kotlow Classification System of Tongue-Tie

# Table 2. NeoEAT Descriptors

NeoEAT Subscale	Purpose	Instrument	Exemplar
Infant Regulation	Infant ability to self- regulate	All	"My baby is satisfied after eating."
Energy & Physiologic Stability	Infant ability to manage the work of eating (suck-swallow-breathe)	All	"My baby is exhausted after eating."
Gastrointestinal Function	Symptoms associated with GI tract dysfunction	All	"My baby gets a bloated (big or hard) tummy after eating."
Oro-Pharyngo-Esophageal Function	Ability to tolerate feeding as it relates to the GI tract	Breast	"My baby chokes or coughs during eating."
Gastroesophageal Function	Symptoms associated with GE disfunction	Breast	"My baby spits up during feeding."
Feeding Efficiency & Sensory Responsiveness	Infant ability to obtain adequate amount of breastmilk with each feeding and sensory components of feeding	Breast	"My baby gets upset when something touches his/her face or mouth."
Sensory Responsiveness	Sensory components of feeding	Bottle & Mixed	"My baby will only eat if fed in a certain way (for example, in a certain chair, or held upright."
Feeding Flexibility	Infant ability to adapt to different modes of feeding (breast and bottle)	Mixed	"My baby prefers breastfeeding over bottle-feeding."
Compelling Symptoms of Problematic Feeding	Serious concerns that indicate feeding problems	Breast & Bottle	"My baby gets pale or blue color around the lips when eating."
Table 3. Demographics of Full Sample (N = 102)

Characteristic	n (%)
Respondent's Relationship to Infant	
Mother	92 (90 2)
Father	9 (8 8)
Other	1(1)
	1 (1)
Respondent's Race	
American Indian/Alaskan Native	1(1)
Asian	2 (2)
Black/African American	1 (1)
White	95 (93.1)
More than 1 Race	2 (2)
Other	1 (1)
Deen on deut?» Ethericiter	
Respondent's Etinicity	2 (2)
Not Hispanie/Latino	2(2)
Not hispanic/Latino	90(88.2)
Unknown	0(3.9)
UIKIIOWII	4 (3.9)
<b>Respondent's Education Level</b>	
Less than high school	2 (2)
High School/GED	16 (15.7)
Associate Degree	18 (17.6)
Bachelor's Degree	31 (30.4)
Master's Degree	28 (27.5)
Doctoral Degree	7 (6.9)
Infant Age	
<pre>&lt; ? months</pre>	79 (77 5)
2 - 4 months	12(11.8)
4 - < 7 months	8 (7.8)
Not Answered	3 (2.9)
1 (ot 7 his worldd	5 (2.9)
Infant Sex	
Male	57 (55.9)
Female	45 (44.1)
Infant Race	
American Indian/Alaskan Native	1(1)
Asian	1(1)
White	86 (84.3)
More than one race	11 (10.8)
Other	2(2)
Not Answered	1(1)
Infant Ethnicity	4 (2,0)
nispanic/Launo	4 (3.9)
Not Hispanic/Latino	00 (00.3) ( (5.0)
Uniter	0 (3.9) 2 (2.0)
UIIKIIOWII Not Answered	5 (2.9) 1 (1)
INOT AIISWEICU	1 (1)

Family Type	
Two-parent	101 (99)
Other	1 (1)
Family Income in USD	
< 15,000	3 (2.9)
15-24,999	2(2)
25-34,999	6 (5.9)
35-49,999	9 (88.2)
50-74,999	16 (15.7)
75-99,999	16(157)
>100,000	48 (47 1)
Unknown	1 (1)
Prior Breastfeeding Experience ( <i>N</i> =62)	
Yes	58 (93 5)
No	4 (6.5)
Dolinow Mode	
Vaginal	01 (70 4)
v aginai C. Saction	81 (79.4)
C-Section	21 (20.6)
Feeding Plan	
Exclusive Breastfeeding	78 (76.5)
Exclusive Formula Feeding	3 (2.9)
Breast and Formula Feeding	4 (3.9)
Exclusive Pumping of Breastmilk	12 (11.8)
Other	2 (2)
Not Answered	2 (2)
Change in Feeding Plans due to Tongue-Ti	e
Yes	25 (24.5)
No	73 (71.6)
Unsure	4 (3.9)
Number of Providers Seen for Tongue-Tie	
Diagnosis	10 (9.8)
0	39 (38.2)
1	30 (29.4)
2	16 (15.7)
3	5 (4.9)
4	2(2)
5 or More	
Infant Medical Diagnoses	
GERD	11 (10.8)
Hearing Impairment	1 (1)
Cardiac Disorder	3(29)
Umbilical Hernia	3(2.7)
Developmental Delay	$\frac{3(2.9)}{1(1)}$
L'aryngomalacia	$\begin{pmatrix} 1 \\ 1 \end{pmatrix}$
Other	1(1)
	2 (2)
Oral Restrictions Diagnosed	
I ongue 11e	102 (100)
Lip Lie	102 (100)

Buccal Tie(s)	35 (34.3)
Tongue-Tie Severity	
II	1(1)
III	92 (90.2)
IV	9 (8.8)
Oral Restrictions Treated	
Tongue Tie	102 (100)
Lip Tie	102 (100)
<b>Complications from Frenotomy</b>	
Yes	0 (0)
No	101 (99)
Unsure	1 (1)
Would you have the tongue-tie corrected	
again? (N= 84)	80 (95.2)
Yes	1 (1.2)
No	2 (2.4)
Unsure	1 (1.2)
Not Answered	

Table 4. Demographics of Subsamples

Characteristic			
	Breastfeeding	Mixed Feeding	<b>Bottle Feeding</b>
	(n = 23)	(n = 27)	(n = 10)
	n (%)	n (%)	n (%)
Respondent's Relationship to Infant			
Mother	21 (91.3)	25 (92.6)	8 (80)
Father	2(8.7)	2(7.4)	1(10)
Other	0(0)	0(0)	1 (10)
Respondent's Race			
American Indian/Alaskan Native	1 (4.3)	0 (0)	0 (0)
Asian	0 (0)	0 (0)	1 (10)
White	22 (95.7)	26 (96.3)	9 (90)
Other	0 (0)	1 (3.7)	0 (0)
To Prove A new			
<pre>Infant Age &lt; 2 months</pre>	22 (95 7)	18 (69 2)	6 (60)
2 - <4 months	1(43)	5 (19.2)	1(10)
4 - < 7 months	0(0)	3 (8.7)	2(20)
			( )
Infant Sex			
Male	12 (52.2)	16 (59.3)	7 (70)
Female	11 (47.8)	11 (40.7)	3 (30)
Infant Race American Indian/Alaskan Native	1 (1 3)	0 (0)	0 (0)
American mulan/Alaskan Native	1(4.3)	0(0)	1(10)
White	21(91.3)	24 (88 9)	7 (70)
More than one race	1(4.3)	1 (3.7)	2(20)
Other	0 (0)	2 (7.4)	0(0)
Family Type			
Two-parent	23 (100)	27 (100)	9 (90)
Other	0 (0)	0 (0)	1 (10)
Family Income in USD	1 (1 2)	1 (2 7)	1 (10)
25-34 000	1(4.3) 2(87)	1(3.7) 1(3.7)	1(10) 2(20)
35-49 999	2(0.7) 1(43)	4(14.8)	2(20) 1(10)
50-74 999	2(8.7)	5 (18.5)	2(20)
75-99.999	3(13)	5 (18.5)	4 (40)
>100,000	14 (60.9)	11 (40.7)	0 (0)
Infant Madical Diagnosas			
GERD	1 (4 3)	4 (14 8)	2 (20)
Hearing Impairment	0(0)	1 (3.7)	0(0)
Umbilical Hernia	0(0)	1 (3.7)	0 (0)
Laryngomalacia	0 (0)	0(0)	1 (10)
Cardiac Disorder	0 (0)	1 (3.7)	1 (10)
Developmental Delay	0 (0)	2 (7.4)	1 (10)
Other	1 (4.3)	1 (3.7)	0 (0)

Oral Restrictions Diagnosed				
Tongue Tie	23 (100)	27 (100)	10 (100)	
Lip Tie	23 (100)	27 (100)	10 (100)	
Buccal Tie(s)	9 (30)	11 (30)	3 (30)	
Tongue-Tie Severity				
II	0 (0)	0 (0)	1 (10)	
III	21 (91.3)	26 (96.3)	6 (60)	
IV	2 (8.7)	1 (3.7)	3 (30)	
<b>Oral Restrictions Treated</b>				
Tongue Tie	23 (100)	27 (100)	10 (100)	
Lip Tie	23 (100)	27 (100)	10 (100)	

Breastfeeding (n = 23)		Mean Score Pre- frenotomy	SD	Mean Score Post- frenotomy	SD
	Total Score	97.1	38.1	69.3*	5.1
	Infant Regulation	16.6	7.4	10.7*	6.3
	Energy & Physiologic Stability	20.4	8.5	14.4*	5.5
	Oro-Pharyngo-Esophageal Function	21.1	8.5	14.5*	6.2
	Gastroesophageal Function	10.3	7.5	8.6	5.6
	Gastrointestinal Function	12.2	6	10	4.6
	Feeding Efficiency & Sensory Responsiveness	13.6	6.4	9*	4.1
	Compelling Symptoms of Problematic Feeding	3	3.4	2	2
Bottle-Feeding $(n = 10)$					
	Total Score	122.8	55.1	75.7*	30.7
	Infant Regulation	17.1	8.8	14.1	7.5
	Energy & Physiologic Stability	26.7	11	14.8*	6.9
	Gastrointestinal Function	66.7	29.4	39.3*	15.3
	Sensory Responsiveness	11.2	8.1	7.3*	6.5
	Compelling Symptoms of Problematic Feeding	1.1	2.1	0.2	0.6
Mixed Feeding $(n = 27)$					
(* 27)	Total Score	122.7	49.8	91.6*	38.4
	Infant Regulation	16.3	6.2	12.9*	4.5
	Energy & Physiologic Stability	23.4	10.9	18*	10.2
	Gastrointestinal Function	53.5	28.3	38*	18.2
	Sensory Responsiveness	9.1	7.7	7.3	7.4
	Feeding Flexibility	20.3	7.6	15.4*	7.4

Table 5. NeoEAT Total and Subscale Scores by Subsample (Aim 1)

\* Indicates significant difference (p < .05) between scores from pre- to post-frenotomy \* These results reflect participants who used the same mode of feeding pre- and post-frenotomy

Breastfeeding		β	t	ρ	95% C	I for $\beta$
					Lower	Upper
	Total Score Change	-1.895	763	.454	-7.060	3.271
	Infant Regulation Change	.130	.263	.795	899	1.159
	Energy & Physiologic Stability Change	204	437	.667	-1.175	.767
	Oro-Pharyngo-Esophageal Function Change	763	-1.176	.253	-2.114	.587
	Gastroesophageal Function Change	362	957	.350	-1.149	.425
	Gastrointestinal Function Change	048	126	.901	835	.740
	Feeding Efficiency & Sensory Responsiveness Change	539	-1.188	.248	-1.482	.404
Bottle- Feeding						
	Total Score Change	-3.6	-2.365	.056	-7.324	.124
	Infant Regulation Change	361	-1.158	.280	-1.080	.358
	Energy & Physiologic Stability Change	066	213	.836	777	.646
	Gastrointestinal Function Change	843	801	.446	-3.270	1.584
	Sensory Responsiveness Change	264	-2.000	.081	569	.040
	Compelling Symptoms of Problematic Feeding Change	.018	.258	.803	144	.180
Mixed Feeding						
	Total Score Change	-1.423	-1.171	.253	-3.924	1.079
	Infant Regulation Change	.309	2.129	.043*	.010	.608
	Energy & Physiologic Stability Change	363	-1.110	.278	-1.036	.311
	Gastrointestinal Function Change	-1.258	-2.052	.051	-2.520	.004
	Sensory Responsiveness Change	.100	.487	.631	323	.524
	Feeding Flexibility Change	211	-1.000	.327	646	.224

# Table 6. Magnitude of Change in NeoEAT Total and Subscale Scores by Infant Age (Aim 2)

\* Indicates significant finding (p < .05)

Symptom	Pre-Frenotomy ( <i>n</i> = 85) <i>n</i> (%)	Post-Frenotomy ( <i>n</i> = 75) <i>n</i> (%)
Creased, cracked, blanching nipples	38 (45)	13 (17)
Painful latching of infant onto breast	61 (72)	21 (28)
Gumming or chewing of the nipples	49 (58)	17 (23)
Bleeding, cracked, or cut nipples	25 (29)	6 (8)
Infant unable to achieve successful latch	50 (59)	6 (8)
Poor or incomplete breast drainage	31 (36)	14 (19)
Infected nipples or breasts	3 (4)	2 (3)
Abraded nipples	9 (11)	1 (1)
Plugged ducts	12 (14)	9 (12)
Mastitis	2 (2)	3 (4)
Nipple thrush	5 (6)	2 (3)
Feelings of depression	8 (9)	2 (3)
Over supply	24 (28)	12 (16)
Under supply	13 (15)	8 (11)
Surgery for breast abscess	1 (1)	0

Table 7. Frequency of Maternal Symptoms Reported in Breastfeeding Mothers

Symptom	β	ρ	95% (	CI for β
			Lower	Upper
Creased, cracked, blanching nipples	3.856	<.001***	1.849	8.042
Painful latching of infant onto breast	6.536	< .001***	3.276	13.040
Gumming or chewing of the nipples	4.644	< .001***	2.327	9.266
Bleeding, cracked, or cut nipples	4.792	.001**	1.842	12.462
Infant unable to achieve successful latch	8.312	< .001***	3.841	17.985
Poor or incomplete breast drainage	2.501	.01*	1.206	5.189
Infected nipples	1.335	.76	.217	8.215
Abraded nipples	8.763	.04*	1.083	70.894
Plugged ducts	1.205	.69	.477	3.044
Mastitis	.578	.56	.094	3.558
Nipple thrush	2.281	.33	.429	12.121
Feelings of depression	3.792	.1	.779	18.452
Over supply	2.066	.07	.949	4.494
Under supply	1.512	.39	.590	3.877
Breast abscess <sup>a</sup>				

Table 8. Logistic Regression of Maternal Symptoms (Aim 3)

<sup>a</sup> Logistic regression could not be run because this symptom was not present in any BF mothers at follow-up.

\* Indicates significant finding (p < .05) \*\* Indicates significant finding (p < .01)

Table 9. Correlation of Maternal Symptoms and NeoEAT Scores Pre-Frenotomy in Exclusively Breastfeeding Mother-Infant dyads (n = 35) (Aim 4)

	NeoEAT Total		Energy & Physiologic Stability		Oro-Pharyngo- Esophageal Function		Feeding E Sensory Re	fficiency & sponsiveness	Compelling Symptoms of Problematic Feeding		
	r	р	r	р	r	р	r	р	r	р	
Maternal Symptom											
Over Supply	.399	.01*	.434	.009**	.387	.02*	.455	.006**	.393	.02*	
Under Supply			.343	.04*							
Poor Latch							.464	.005**			

\* Indicates significant finding (p < .05) \*\* Indicates significant finding (p < .01)

\*\*\* Indicates significant finding (p < .001)

Table 10. Correlation of Maternal Symptoms and NeoEAT Scores Post-Frenotomy in Exclusively Breastfeeding Mother-Infant dyads (n = 39) (Aim 4)

	NeoEAT Total		NeoEAT Total		NeoEAT Infant Energy & Total Regulation Physiologic Stability		ergy & siologic ability	Gastrointestinal Function		Oro- Pharyngo- Esophageal Function		Feeding Efficiency & Sensory Responsiveness		Compelling Symptoms of Problematic Feeding	
	r	р	r	р	r	р	r	р	r	р	r	р	r	р	
Maternal Symptom															
Painful Latch	.366	.02*	.374	.02*	.340	.03*	.332	.03*			.379	.01*	.364	.02*	
Poor Latch	.486	.002**			.416	.008**	.405	.01*			.472	.002**	.353	.02*	
Creased/Cracked Nipples	.367	.02*			.362	.02*	.327	.04*			.475	.002**			
Gumming/Chewing on Nipples	.426	.007**	.535	<.001***	.405	.01*	.323	.05*			.430	.006**	.382	.02*	
Bleeding Nipples											.386	.02*	.339	.04*	
Incomplete Drainage	.413	.009**	.479	.002**					.354	.03*	.337	.04*	.482	.002**	

\* Indicates significant finding (*p* < .05) \*\* Indicates significant finding (*p* < .01)

\*\*\* Indicates significant finding (p < .001)

## References

## **Refer to Cumulative Reference List**

# Chapter V

## Summary of the dissertation

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#### Discussion

Tongue-tie is a common, understudied oral anomaly that has the potential to interfere with infant feeding (Hill & Pados, 2020). While treatment has not demonstrated severe complications, it remains controversial. Unfortunately, healthcare providers differ in their beliefs and understanding of how tongue-tie may complicate feeding (Walsh, Links, Boss, & Tunkel, 2017). Nonetheless, recognition and treatment of tongue-tie has increased dramatically over the last decade (Walsh & Tunkel, 2017). Given the emphasis for exclusive breastfeeding for the first six months of life by the WHO (World Health Organization, 2019), CDC (Office of Disease Prevention and Health Promotion, 2019), and UNICEF (2015), it is important to critically evaluate how tongue-tie may lead to early cessation of breastfeeding, and maternal or infant symptoms that occur due to a tight sublingual frenulum.

As such, the purpose of this program of research was to evaluate the prevalence of tongue-tie in the infant population, broadly examine the state of the science of tongue-tie, and explore how tongue-tie and its treatment contribute to both maternal and infant symptoms associated with feeding. Cumulatively, this program of research has made incremental and meaningful contributions to the literature about tongue-tie, with implications for future research that will aid in an improved understanding of the anomaly, what mother-infant dyads benefit greatest from tongue-tie correction, and additional physiologic factors affected by tongue-tie.

This discussion presents a summary of the dissertation findings in association with previous research, centered on three main areas: 1) prevalence of tongue-tie in the infant population, 2) evaluation of tongue-tie, and 3) symptoms of problematic feeding for both mothers and infants in the setting of tongue-tie. Following this discussion, theoretical,

methodological and clinical implications are summarized. Areas for future research are also recommended based on these three main themes.

## **Prevalence of Tongue-Tie**

With the recognition that prevalence reporting has been highly variable between studies, meta-analytic approaches were used to determine a more accurate estimate of tongue-tie prevalence in the population of children less than one year of age in Chapter II. Fifteen studies were included in the meta-analysis and overall prevalence of tongue-tie was determined to be 8%. Prevalence was higher in males and when a standardized assessment was used to make a diagnosis, although neither finding was statistically signifficant. The assessment tools used to diagnose tongue-tie did not have psychometric testing to support use, the risk of bias in the 15 studies was moderate to high, and some did not include an acceptable case definition to confirm a diagnosis of tongue-tie. The prevalence rate of 8% falls within the range reported in the literature (Cetinkaya et al., 2011; Ngerncham et al., 2013). The increase in prevalence in male infants is consistent with our understanding of the sex-linked genetic component (Hazelbaker, 2010) and the findings from previous research that compared prevalence by sex (De Oliveria et al., 2019; Flinck et al., 1994; Freudenberger et al., 2008; Perez-Aguirre et al., 2018). Within Chapter II, significant advancements were made to better understand how common tongue-tie is in the infant population with an understanding that more work is needed to allow for consistent diagnosis and comparison within and between studies.

## **Evaluation of Tongue-Tie**

Chapters II and III critiqued how evaluation of tongue-tie is performed and how a diagnosis is made. Consistently throughout both chapters, the diagnosis of tongue-tie was

made subjectively or using a diagnostic tool that has not been tested psychometrically (Hill & Pados, 2020; Hill, Lee & Pados, 2020). This is an important consideration when reviewing all results, and until the available tools are evaluated for reliability and validity, this will remain a limitation of current and future research. In both chapters II and III, the differences between classification systems, the method(s) used to diagnose tongue-tie, and the indications for when tongue-tie may be suspected were explored. It became evident that there is such great variability in these processes that it is not possible to determine best practice guidelines for screening or diagnosis or to fully analyze results between studies. The research is also inconsistent in the dyadic evaluation; some studies evaluate only the mother, others focus solely on the infant. None of the research published to date has provided a comprehensive evaluation of both the mother and the infant to help guide interventions or treatment. In addition, most studies focus on breastfeeding infants, and we know that the majority of infants are bottle fed; thus, we are missing evalution of tongue-tie in bottle-fed infants. Chapter IV did address this, but the sample of bottle-fed infants was very small (n = 10) and thus, not a true or generalizable representation of the population.

A limitation in Chapter IV and the research critiqued to date is that matched control groups or participants with untreated tongue-tie have not been included, especially over the long term. In a small number of studies that attempted to include a control group (Buryk et al., 2011; Emond et al., 2014; Hogan, Westcott, & Griffiths, 2005), infants that were diagnosed with tongue-tie whose caregiver elected not to undergo frenotomy then chose to have the procedure within 2 weeks of data collection. This has resulted in an inability to evaluate untreated tongue- tie and its effect on feeding for both mother and infant longitudinally. It does provide data to suggest that untreated tongue-tie resulted in persistent

feeding challenges, which is why caregivers pursued treatment shortly after the studies began. In the critically important area of infant feeding it would be unethical to withhold treatment for research purposes, and this remains a challenge in designing a randomized control trial in this population.

## Symptoms of Problematic Feeding in the Setting of Tongue-Tie

Symptoms of problematic feeding for infants with tongue-tie was critically examined in both Chapters III and IV. In Chapter III, a systematic review determined that frenotomy is most beneficial when performed within the first month of life (Hill & Pados, 2020). Frenotomy was found to be a safe and minimally-invasive procedure. The literature synthesized focused largely on maternal symptoms and the improvement of those symptoms following frenotomy, mostly nipple pain and LATCH scores. The systematic review revealed significant limitations. First, anatomical assessments of infants was variable between studies, making comparison impossible. Second, maternal anatomy was not considered as a component of breastfeeding evaluation, which limits our ability to understand how maternal anatomy in addition to tongue-tie may alter feeding. Third, in the studies that did include infant symptoms (i.e., suck quality and milk transfer), the sample sizes were small and inadequately powered (Geddes et al., 2008). Lastly, none of the published research evaluated infant feeding or symptoms of problematic feeding using a validated, comprehensive measure (Hill & Pados, 2020).

To address some of these limitations, a study was designed and implemented in Chapter IV. A single-site was utilized for participant recruitment, allowing for consistency in tonguetie diagnosis, with inclusion of both maternal and infant symptoms and the use of a validated and comprehensive evaluation of infant feeding. Both maternal and infant symptoms were examined along with tongue-tie severity using Kotlow's diagnostic criteria (Kotlow, 1999). This study was the first to examine both maternal and infant symptoms and evaluate them using a variety of statistical methods to help us better understand maternal and infant symptoms that are associated with tongue-tie, how these symptoms improve following frenotomy, and describe problematic infant feeding using a validated assessment measure.

Consistent with previous knowledge described in Chapter I, results from the study in Chapter IV support the explanation as to how tongue-tie causes difficulties with breastfeeding. As previously explained by Genna (2017), when the tongue cannot extend below the lower gumline, reflexive biting may occur. Biting or gumming on the breast nipple was reported by 49 of the 85 breastfeeding women prior to frenotomy (58%), and was correlated with painful latch. A physical assessment of the infant or mother did not occur in this study, so we cannot determine if this was associated with tongue extension in our sample. An infant's inability to accept the breast nipple not only reduces latch ability, it has also been noted to create friction and pain for the breastfeeding mother (Genna, 2017). Mothers in our study most commonly presented with painful latch (n = 61, 72%), but an evaluation of a breastfeeding session was not performed to determine if changes in position or other lactation support may have relieved this symptom. Poor latch, painful latching and chewing/gumming on the breast nipple were all significantly more likely to be present prior to frenotomy and these symptoms were markedly reduced after frenotomy in the breastfeeding subsample. Reports of bleeding nipples, oversupply of breastmilk, or nipple thrush pre-frenotomy were related to significantly reduced NeoEAT scores post-frenotomy. This knowledge provides valuable information for parents and providers on which maternal symptoms are most associated with problematic symptoms of feeding for the tongue-tied infant.

Advancements made in tongue-tie research. Across these three chapters (II, III, IV), significant advancements in the understanding of tongue-tie were made. First, a more accurate prevalence of the anomaly was determined. Next, symptoms that have been previously evaluated were described and critiqued. Maternal symptoms that contributed to problematic feeding were identified and included in a systematic and thoughtful way in Chapter IV. To highlight the major differences in this work comprared with previous studies, it is the first to comprensively evaluate infant feeding and associated problems both prior to and following frenotomy.

With the understanding that feeding is a complex process, with changes over time as the infant practices and matures, comparison of infant feeding and problematic symptoms were evaluated over a short period of time. This was done intentionally to limit the potential that growth and maturation contributed to changes in symptoms. The influence of tongue tie and its correction on feeding was also examined with a moderating effect of infant age, as we know that feeding transitions from a reflexive reflex response to voluntary feeding around four months of age (Glodowski, Thompson, & Martel, 2019) and improvements in feeding symptoms may vary in older infants as the child has more time to practice feeding with their natural anatomy.

## **Summary and Implications**

**Theoretical implications.** The cumulative results of this body of work supports and adds to Lenz's Theory of Unpleasant Symptoms (Lenz, Pugh, Milligan, Gift & Suppe, 1997) (Figure 1). This work identified evidence of particular maternal symptoms that correspond with more problematic infant feeding. In Chapter III, we reviewed the published literature, noting that the majority of research published to date considers only one aspect of the

influence of tongue-tie and its correction -- maternal symptoms and their improvement. Namely, nipple pain was the most commonly researched maternal symptom, while infant symptoms had not been studied to determine efficacy of frenotomy to treat tongue-tie (Hill & Pados, 2020). The combination of both maternal and infant symptoms is an important consideration in the determination of who may benefit most from frenotomy to correct tongue-tie.

In Chapter IV, the assessment of both maternal and infant symptoms effectively demonstrated how physiologic factors in both members of the feeding dyad affect the performance activity of interest, infant feeding. For infants who bottle-feed, situational factors, such as bottle and/or nipple type are important considerations that may alter feeding performance, especially in infants with tongue-tie. Without accounting for these situational factors, bottle-fed infants demonstrated a significant improvement in problematic feeding after frenotomy.

In breastfeeding dyads, maternal and infant symptoms were associated with tongue-tie, and they improved following frenotomy. Infants demonstrated significant improvements in symptoms of problematic feeding related to nearly all aspects evaluated. Frenotomy improved infant ability to self-regulate, conserve energy, improve efficiency with feeding, and reduce gastroesophageal and gastrointestinal symptoms. The symptoms reported by breastfeeding mothers ranged in frequency; however, all symptoms (e.g., painful or poor latch, chewing on the nipple) were reduced following tongue-tie correction. Most notably, when the tongue-tie was corrected and the infant and maternal symptoms improved, infant feeding performance improved enough so that mothers were able to exclusively breastfeed or increase breastfeeding when they had previously been unable to do so. These factors have been applied to Lenz's theory to help theoretically explain how physiologic factors in both mother and infant create symptoms that affect feeding performance in infants with severe tongue-tie (Figures 2 and 3). This theory can be utilized to inform future work in this area. Situational and psychologic factors should be considered as additional causes of unpleasant symptoms with feeding for infants with tongue-tie. Figure 1. Original Figure/Theory







Figure 3. Infant Symptoms



Methodological implications. This dissertation builds on well-validated measures of problematic infant feeding, the NeoEAT instruments (Pados, Thoyre, Estrem, Park, & McCormish, 2018; Pados et al., 2019a; Pados, Thoyre, & Galer, 2019), to better identify and understand symptoms of problematic infant feeding and how those symptoms may improve following treatment of tongue-tie. There were significant improvements in many of the NeoEAT subscales for infants fed by breast, bottle, or a combination of the two. These findings support continued use of the NeoEAT measures to evaluate infant feeding in conjunction with oral assessment for tongue-tie. Maternal symptoms of oversupply, undersupply, and painful latch while breastfeeding were significantly correlated with NeoEAT total and subscale scores. Despite a decrease in maternal symptoms after frenotomy, in cases where symptoms persisted there were significantly higher NeoEAT total and subscale scores. This suggests a strong association between maternal and infant symptoms as they relate to difficulties with feeding. In addition, even when tongue-tie is no longer present, the NeoEAT measures can be helpful in idenfying the association between continued infant symptoms that occur while breastfeeding.

**Clinical implications.** The purpose of this dissertation was to develop a better understanding of the prevalence, available research, and symptoms associated with tongue-tie. This program of research has great utility in clinical practice. Having a better understanding of the prevalence of tongue-tie, the symptoms for both mothers and infants that lead to more problematic feeding, and the improvements in those symptoms post-frenotomy will help clinicians better appreciate the physiologic implications of tongue-tie. Additionally, a dyadic approach is warranted when evaluating breastfeeding, which is an important component of holistic nursing care. This body of works supports not only assessing for maternal symptoms associated with feeding challenges, but also taking into account the specific infant symptoms that are occurring simultaneously. The results in Chapter IV also support the use of the NeoEAT measures to evaluate infant feeding in the setting of tongue-tie.

As discussed in the introduction of this dissertation (Chapter 1), there have been studies that suggest early cessation of breastfeeding for babies with tongue-tie (Ricke, Baker, Madlon- Kay & DeFor, 2005; Kent et al., 2015). While more than 80% of mothers initiate breastfeeding, far less achieve EBF for the first six months of life (Centers for Disease Control and Prevention, 2018). The data from participants in our study in Chapter IV support these findings. Nearly all (98%) participants had a feeding plan prior to the birth of the infant, and 78 (78%) reported the feeding plan was to exclusively breastfeed at the breast. However, only 35 (34%) of those respondents were actually exclusively breastfeeding at the time of the study. Moreover, 25 (24.5%) participants stated that their baby's tongue-tie altered their initial feeding plan. These are important findings that need to be explored in greater depth. If tongue-tie is impeding breastfeeding success and reducing rates of EBF, strategies to support the breastfeeding dyad should be developed. Knowing now that tongue-tie affects 8% of the infant population and that it may have significant implications for the development of problematic feeding symptoms supports research and practice priorities focused on assessment, diagnosis, and intervention strategies.

**Strengths.** This body of work has several strengths. First, it represents the first metaanalysis of prevalence data to determine a more accurate prevalence rate of tongue-tie for use in future research and clinical practice. Second, it outlines the major gaps in the research to date, with the recognition that infant feeding and symptoms of problematic feeding have not been addressed in the available research. Knowing that, we selected robust, well-validated measures to evaluate infant feeding in infants with tongue-tie before and after correction. A variety of statistical approaches were utilized to critique and analyze data, including metaanalytics, general linear modeling, linear and logistic regression, correlations, and t-tests as appropriate to advance the state of the science of tongue-tie. The data presented in each of the manuscripts was organized clearly and succinctly, allowing for use in clinical practice while simultaneously including limitations and recommendations for future research.

**Limitations.** It is important to note the limitations of this manuscript dissertation. While the meta-analysis included a large sample to analyze prevalence data (N = 24,536). there was significant heterogenieity within and between studies. Future work is needed to improve the operational and conceptual definitions of tongue-tie that will aid in an improved ability to evaluate prevalence data in future research. Data from Chapter IV was limited to a sample of participants from a single practice site in the northeast region of the United States, which was homogenous in race and geographic location. This sample had little variation in tongue-tie severity. Thus, findings are limited to this subgroup. The diagnostic criteria to determine tongue- tie severity used in that same study has not been psychometrically evaluated. The evaluation of maternal symptoms was based on mother's subjective report, as a valid and reliable measure to assess these symptoms does not currently exist. While data was collected at two time points, longitudinal studies will better explore and explain how tongue-tie impacts feeding. The research on tongue-tie to date is limited by inconsistent definition and measurement of the anomaly and the symptoms that may ensue. This program of research made efforts to define and address those inconsistencies through the systematic review of the literature that was then used to design the study in Chapter IV.

## **Future Research**

This program of research is a forward-step to a better understanding of the efficacy of tongue-tie treatment, though replication of this research in larger, heterogenous samples is needed. While the area of interest was tongue-tie, all infants were also concurrently treated for lip-tie, and this anomaly needs to be explored further. In addition, physiologic data to evaluate changes in sucking patterns and suck strength will provide information on the utility of frenotomy to improve suck mechanics and feeding efficiency. Despite the methodological rigor and findings from this work, additional research is necessary to better understand prevalence of tongue-tie, explore the utility of the screening tools available for use, and improve our knowledge of the symptoms that both mother and infant display when this anomaly is present.

**Tongue-Tie Prevalence.** Understanding that prevalence was highly variable between these studies, psychometric evaluation for diagnostic tools is missing, and the variation in how each study defined tongue-tie are important findings to guide future research. There remains a need for the development of a universal diagnostic measure that incorporates the newly revised definition of tongue-tie (Messner et al., 2020). Evaluation and input from multiple disciplines (e.g., lactation counselors and practitioners) is needed to understand how each specialty provider evaluates the mother-baby dyad and makes a tongue-tie diagnosis. Overall, the study of tongue- tie in infants has been limited by the absence of a universally accepted definition, lack of valid and reliable assessment measures, and suboptimal evaluation of problematic feeding symptoms in the infant (O Shea et al., 2017; Hill & Pados, 2020). Once these limitations are addressed, a large study is needed to determine the true prevalence of tongue-tie in infants and factors that may be associated with this anomaly.

**Tongue-Tie Screening and Diagnosis.** Throughout this dissertation, it is evident that the methods to screen for and diagnose tongue-tie varies between providers and research

studies.

The guidelines for what constitutes a diagnosis of tongue-tie and its revision has demonstrated marked heterogenieity within and between studies. With that, the diagnostic measures available for use have not been evaluated psychometrically. Knowing this, future research must aim to develop consistency of assessment procedures and critique the assessment tools used in practice. From there, research to determine critiera for treating tongue-tie can be developed. If the current measures are not psychometrically sound, additional research will be necessary to create a valid and reliable instrument that demonstrates inter-rater agreement over time. These advancements will allow researchers, clinicians, and parents to compare findings between studies and move us closer to a universal screening recommendation.

**Symptoms of Problematic Feeding.** The understanding of tongue-tie and the complications of this anomaly on infant feeding is necessary for several reasons. First, while a consensus agreement was recently published (Messner, 2020), it recognizes that tongue-tie may be a cause for breastfeeding problems, namely nipple pain. A better understanding of how tongue-tie affects infants and their ability to successfully feed, regardless of feeding mode, will improve our understanding of when the anomaly should be treated.

In Chapter IV, the samples included in each subgroup of breastfeeding, bottle-feeding and mixed feeding were small, as were the samples in the 2 - < 4 month and 4 - < 7 month age groups. Larger studies should be conducted to evaluate infant symptoms in a more diverse and representative sample of the population. Maternal symptoms were obtained by self-report; examination of maternal anatomy in future studies would add to the research to help identify other factors that may be causing these negative symptoms. For example, flat nipples are frequently associated with poor latch and pain with breastfeeding (Dewey, Nommsen-rivers, Heinig, & Cohen, 2003; Kent et al., 2015), and is unrelated to tongue-tie. The continued report of maternal symptoms following frenotomy indicates that there may be other causes of these symptoms. Alternatively, there may be some mother-infant dyads who need more time to adjust to feeding after frenotomy. Most of the symptom analyses from Chapter IV focused on breastfeeding mother-baby dyads. Infant symptoms as they relate to bottle feeding and as these infants transition to solid foods later in life will provide additional data to understand the effects that tongue-tie has on feeding over time.

Longitudinal research that includes comprehensive breastfeeding evaluation would help us better understand factors that lead to continued maternal symptoms such as painful latch or oversupply. Since we do not know how many infants did not have a diagnosis of tongue-tie or which dyads elected not to have tongue-tie corrected, future research that includes both a norm- reference group and a sample of infants with tongue-tie that is not corrected would strengthen our knowledge on how tongue-tie impacts feeding. As feeding is directly related to growth and development, evaluation of weight gain and the successful achievement of developmental milestones over time would be useful variables to measure in future work.

## Conclusions

In summary, this program of research has made substantial and meaningful contributions to the science of tongue-tie, its prevalence, and associated maternal and infant symptoms. The body of work presented in this dissertation representes advances in this area, in conjunction with theoretical, methodological, and clinical implications. There is a need for future research to develop a greater understanding of how to best screen for tongue-tie, to determine if tongue-tie severity impacts symptom improvement for both the mother and baby,

the physiologic implications of the anomaly, and our clinical management of tongue-tie.

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