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Standardizing Assessment and Management of Ankyloglossia

by

Marcia Cowen

A Scholarly Project Submitted to the College of Nursing in Partial Fulfillment of the
Requirements for the Degree
Doctorate of Nursing Practice

University of New Mexico

College of Nursing

Albuquerque, New Mexico

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“Standardizing Assessment and Management of Ankyloglossia”

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Abstract

Ankyloglossia, which is characterized by a short, tight or thick lingual frenulum, can negatively impact breastfeeding. It has been recognized as a problem for centuries. Frenotomies have traditionally been performed to remove the lingual frenulum when indicated. The use of frenotomies and a focus on ankyloglossia fell out of favor in the twentieth century with renewed attention in the past several decades. Limited high-quality studies have been undertaken on ankyloglossia. There remains no standardized diagnosis or management recommendations. In efforts to promote a standardized approach that is based on current knowledge, this study will involve an educational in-service on ankyloglossia, and the introduction of an objective assessment tool with associated recommendations for management of ankyloglossia within a pediatric primary care setting. Additionally, provider perceptions of ankyloglossia will be explored.

Keywords: ankyloglossia, tongue-tie, breastfeeding, frenotomy, lingual frenulum

Dedication

I am very thankful for the support of my husband Dr. Benjamin Cowen. We have supported each other through numerous years of higher education, pursuing our professional goals, dreams, our faith and beginning our family together. I am honored by the families who have allowed me to care for them when they are most vulnerable and trust me with the care of their little ones. My professional nursing experience first sparked my interest in ankyloglossia. My experience following the births of my children along with the recommendations I received about breastfeeding and ankyloglossia encouraged me to explore the topic further; desiring to be able to provide families with knowledgeable and thoughtful advice. Finally, I dedicate this scholarly project to my sons, Graham, Zachary, and Luke. You have kept me grounded throughout this whole process and inspire me daily.

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List of Acronyms

ABM: Academy of Breastfeeding Medicine

BBAT: Bristol Breastfeeding Assessment Tool

BSES-SF: Breastfeeding Self-Efficacy Scale- Short Form

BTAT: Bristol Tongue Assessment Tool

HATLFF: Hazelbaker Assessment Tool for Lingual Frenulum Function

IBFAT: Infant Breastfeeding Assessment Tool

I-GERQ-R: revised infant gastroesophageal reflux questionnaire

LATCH: latch, audible swallowing, type of nipple, comfort, and hold-tool

RCTs: randomized controlled trials

SF-MPQ: Short-Form McGill Pain Questionnaire

VAS: visual analogue scale

WHO: World Health Organization

Introduction and Background

Ankyloglossia, which is also known as tongue-tie, is a condition characterized by a short, tight or thick lingual frenulum (O’Shea et al., 2017). It has been associated with breastfeeding difficulties, including poor latch, nipple pain, and suboptimal weight gain (Merrit, 2019).

Approximately 25-44% of infants with ankyloglossia will have feeding difficulties (Ingram et al., 2015). Traditionally, frenotomies have been the treatment. The procedure involves removing the lingual frenulum. The practice of frenotomies had fallen out of favor as bottle feeding became common place (Steehler et al., 2012). Recent resurgence of the practice is thought to be secondary to an increased focus on breastfeeding (Walsh et al., 2017). Ankyloglossia remains poorly understood with conflicting evidence about proper assessment, management and outcomes for babies and breastfeeding moms (Francis et al., 2015).

There is no agreed upon definition of ankyloglossia. The International Affiliation of Tongue-tie Professionals (n.d.) has created their own definition, “the lingual frenulum (or frenum), is a remnant of tissue in the midline between the undersurface of the tongue and the floor of the mouth. When it interferes with normal tongue function it is called ‘symptomatic tongue-tie’ or ‘symptomatic ankyloglossia’”. Lingual frenulums are thought to be a remnant from embryological development. For most infants they disappear before birth secondary to apoptosis, which is the programmed cell death of specific cells (Hazelbaker, 2010). It is unknown why they remain for some. Ankyloglossia is estimated to occur in 0.1-12.11% of infants, although without agreed upon diagnostic criteria, exact prevalence rates are unknown (Walsh & Tunkel, 2017).

While ankyloglossia has received much attention in recent years, it is not a new concept. Some argue that it is referenced in the Old Testament by Moses who states he is “slow of speech and of tongue” (Exodus 4:10b, ESV; Kotlow, 2011). Ancient Greeks discussed surgical

management for tight frenulums and there is evidence that centuries later during the Middle Ages, frenotomies were still being performed (Obladen, 2010). Both midwives and physicians commonly performed frenotomies until the twentieth century; the procedure became less common as formula feeding became the norm in the 1940s and 1950s (Hogan et al., 2005; Muldoon et al., 2017; Steehler et al., 2012). Frenotomies continued to decrease in the late twentieth century (Ingram et al., 2015). As the maternal and infant health benefits of breastfeeding were realized starting in the 1980s and 1990s, there was a gradual increased focus on breastfeeding and an increased focus on ankyloglossia (Steehler et al., 2012).

Efforts to promote breastfeeding have been far reaching. The World Health Organization (WHO) published their Ten Steps to Successful Breastfeeding in 1991 and a revised version was published in 2018 (Baby Milk Action, 2019). The Baby-Friendly Hospital Initiative is a global initiative that was created in 1991 to promote breastfeeding, and incorporated the WHO's Ten Steps to Successful Breastfeeding (WHO, 2019). The Academy of Breastfeeding Medicine was established in 1993 (ABM, 2019). The Office of the Surgeon General published their breastfeeding Call to Action in 2011, in which they outline strategies to promote exclusive breastfeeding (U.S. DHHS, 2011). With all these efforts, rates of breastfeeding have gradually increased. Rates of exclusive breastfeeding at six months of age within the United States increased from 16% to 25% from 2009 to 2016 (CDC, 2019).

Diagnosis and treatment of ankyloglossia with frenotomies has been increasing in recent years. A 2017 retrospective audit study found that the number of in-patient cases of ankyloglossia diagnosed from 1997 to 2012 within the United States increased from 3,934 to 32,837. Not surprisingly, there was also an increase in frenotomy procedures for the same timeframe, increasing from 1,279 to 12,406 (Walsh et al., 2017).

Problem Statement

While frenotomies for the management of ankyloglossia have been on the rise, there remain no universally accepted standards for assessment and management of ankyloglossia for newborns. Additionally, few high-quality studies have been conducted to guide clinical decisions related to ankyloglossia. Much of the literature available has been plagued by methodological flaws. Breastfeeding is well supported as the ideal nutrition for infants and has maternal and infant benefits. Ankyloglossia has been reported to negatively impact breastfeeding for some mother/infant dyads. Not all infants with lingual frenulums will have feeding difficulties, and therefore frenotomies are not needed for all cases of ankyloglossia. Currently the assessment and management of ankyloglossia is variable, and likely frenotomies are over-performed for some and not offered to others who might benefit from the procedure. A standardized approach to the diagnosis and management of ankyloglossia is needed to minimize unnecessary frenotomies while simultaneously recommending them for the infants most likely to have beneficial outcomes.

PICOT Question

In providers caring for newborns in an ambulatory care setting in an urban area, does an educational in-service and the use of an objective grading tool for ankyloglossia compared to subjective assessment for ankyloglossia lead to a more standardized approach for clinical management of ankyloglossia and/or any change in provider perception of ankyloglossia within six weeks?

Objective and Aims

The objectives and aims of this project are as follows:

1. Perform an educational in-service for providers and nurses within an ambulatory pediatric clinic addressing what is currently known about ankyloglossia

2. Introduce the use of an objective measurement tool for ankyloglossia
3. Standardize the assessment and management of ankyloglossia within a pediatric setting
4. Explore provider perception of ankyloglossia through questionnaires administered pre/post educational in-service

Review of Literature

Ankyloglossia has been the focus of much attention in recent years both within the medical community and by families. The heightened focus has been mainly related to ankyloglossia's connection to breastfeeding and recognition of breastmilk as the ideal nutrition for babies. Ankyloglossia, which is a shortened, tight or thick lingual frenulum, is still poorly understood, with conflicting evidence about proper assessment, management and outcomes for babies and breastfeeding moms (Francis et al., 2015). Additionally, many of the studies on ankyloglossia have had small sample sizes, with the total number of infants amongst all groups numbering thirty to fifty, with only short-term outcomes being assessed retrospectively, thus further limiting their reliability and generalizability.

The main treatment for ankyloglossia has traditionally been frenotomies. Frenotomies can be done in a simple procedure using sterile scissors or via laser to reduce the tight frenulum. For mother/infant dyads with breastfeeding difficulties, reported benefits have included decreased maternal pain, increased milk supply, improved weight gain and continued breastfeeding for a longer duration (Brookes & Boweley, 2014).

With conflicting evidence regarding ankyloglossia, there is controversy within the medical community about whether frenotomies are warranted and what impact ankyloglossia plays in impacting breastfeeding. Providers' personal thoughts on ankyloglossia drastically shape their recommendations for families.

This review will investigate what is known and what is still lacking in knowledge about ankyloglossia. PubMed and CINAHL were searched using the key terms of ankyloglossia, tongue-tie, frenotomy and breastfeeding. The terms ankyloglossia, tongue-tie and frenotomy were searched individually and in combination with “and breastfeeding”. No limits were set on year of publication due to the limited total number of studies. Articles pertaining to ankyloglossia and speech difficulties or that did not pertain to infants were excluded. Upon reviewing the literature, it became evident that there are five randomized controlled trials (RCTs) that have been done to date on ankyloglossia that are cardinal articles. Additionally, studies have explored posterior ankyloglossia, labial frenulums, provider perception, ideal timing of frenotomies, frenotomy revisions, and development of assessment tools. The five RCTs and most of the current studies on ankyloglossia are reviewed below. Studies were selected to cover ankyloglossia comprehensively, to ensure addressing current research on the topic while covering aspects that are applicable for this scholarly project.

Objective Assessment Tools for Ankyloglossia

Kotlow developed the Kotlow classification system for tongue-tie in infants in 2004 and created a revised version in 2011 for children 2.5-5 years of age (Kotlow, 2011). Four classes are identified, based on where the lingual frenulum attaches to the tongue, with Class I being closest to the tip of the tongue and Class IV being towards the posterior aspect of the tongue (Kotlow, 2004). Classes I and II are considered anterior ankyloglossia while Classes III and IV are considered posterior. According to Kotlow (2011), classes II, III, and IV may hinder breastfeeding and those infants should be assessed for possible frenotomies. Similarly, Coryllos et al., (2004) developed a classification system for ankyloglossia based on location of attachment of the lingual frenulum to the ventral surface of the tongue. The Coryllos classification system

also has four classes with classes I and II being considered anterior and classes III and IV being posterior. The main difference between the Kotlow and Coryllos systems are the precise measurements that differentiate the sub types. Both the Kotlow and Coryllos systems are based on anatomical findings alone and do not address function of the tongue or frenulum.

The Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) was originally created in 1993 by Dr. Alison Hazelbaker as part of her Master's thesis and then revised in 2010 (Hazelbaker, 2010). The tool has five appearance items and seven functional items, each which may receive a score of 0-2 with total possible scores ranging from 0-24. Items include appearance of tongue when lifted, elasticity of frenulum, length of lingual frenulum when tongue lifted, attachment of frenulum to tongue and inferior alveolar ridge, lateralization, lift of tongue, extension of tongue, spread of anterior tongue, cupping, peristalsis and snapback (Hazelbaker, 2010). Frenotomies are recommended if the functional score is 10 or less, or if the appearance score is 7 or lower. Regardless of appearance scores, if the functional score is higher than 13 frenotomies are not recommended. If the functional score is 11-13 with an appearance score greater than 9, frenotomies are not recommended (Hazelbaker, 2010). This tool has been difficult to incorporate into practice due to its complexity. The length and elasticity of the frenulum items have been the most challenging to achieve consistent reliability (Ingram et al., 2015).

Recognizing the limitations of previous assessment tools, Ingram et al. (2015) developed the Bristol Tongue Assessment Tool (BTAT). They sought to develop a tool that was simple, easy to use, assessed function and allowed for objective recommendations pertaining to whether frenotomies should be offered. Three midwives determined four characteristics they deemed most significant for assessing ankyloglossia based on previous assessment tools and their

clinical experience. The four characteristics were tongue tip appearance, attachment to the lower gum ridge, lift of the tongue and protrusion. Each characteristic may receive a score of 0-2, with a total score ranging from 0-8. Scores 0-3 indicated the most severe impairment from ankyloglossia. To further validate the tool, it was compared to the HATLFF and underwent a process for internal consistency as well. Thirty-three infants were assessed by two separate midwives with the BTAT and scores were compared. The reliability of the tool was acceptable with a Cronbach's alpha score of 0.708. Additionally, 224 infants were assessed with the BTAT and 126 with the HATLFF to determine whether the tools were comparable. The Pearson correlation test showed a strong correlation between the two tools (Pearson=0.89, $p < 0.001$). Midwives found the BTAT simpler to use and understand. Ingram et al. (2015) made a strong case for introducing the BTAT as the ideal objective measure for function associated with ankyloglossia.

Randomized Controlled Trials

The first RCT on ankyloglossia was conducted in 2005 by Hogan et al. They sought to determine whether lactation support or frenotomy produced greater improvement on infant feeding for infants with ankyloglossia and feeding difficulties. Their study included bottle fed and breastfed babies. Feeding difficulties for breastfed babies were defined as poor latch, nipple pain, or continuous feeding, and for bottle fed babies as poor latch, slow speed of feed, dribble and sucking in excess air. Within the sample, 201 infants had ankyloglossia, 88 of whom had feeding issues (75 breastfed vs. 13 bottle). Thirty-one were not enrolled due to parents wanting immediate frenotomies, improvement in breastfeeding within four weeks, or switching to bottle feeding or feeding without difficulty at four weeks. Therefore 57 (40 breastfed vs 17 bottle) were randomized; 29 to receive 48 hour lactation support and 28 to immediate frenotomy). The average age at frenotomy was 20 days of life although it varied from 3-70 days. Follow up was

completed via telephone at twenty-four hours post intervention and then weekly for four weeks and once again at four months. The control group was offered frenotomies after forty-eight hours. Feeding outcomes were solely based off of maternal report.

One control improved with lactation support alone. The remaining 27 all chose to receive frenotomies after forty-eight hours of lactation support. All but one from the intervention group had improvements in feeding post frenotomy. The overall improvement rate after frenotomy, including all infants who received the procedure, was 97% (54/57, $p < .001$). Breastfed babies randomized to the intervention group were found to have a 60% breastfeeding rate at four months follow up. Hogan et al. (2005) also reported no complications from frenotomies, that feeding improvements tended to be immediate, and that there was no relationship between degree of ankyloglossia and degree of impairment on infant feeding. Major limitations included no blinding, 54/57 infants ultimately received frenotomies limiting longer term outcome assessment, overall small sample size, and no objective measures assessing infant feeding or severity of ankyloglossia.

Dollberg et al. (2006) sought to build upon the Hogan study. The purpose of their study was to assess whether frenotomy affected infant LATCH scores and maternal nipple pain using the standard visual analogue pain scale. The LATCH tool assesses infant latch while breastfeeding, and assesses five areas: latch, audible swallowing, type of nipple, maternal comfort, and need to hold the baby to the breast. It assigns a score of zero to two for each parameter. Twenty-five infants with ankyloglossia whose mothers had nipple pain were recruited from one lactation clinic and randomized to have a frenotomy ($n=14$) or a sham procedure ($n=11$). The study was double blinded; researchers interacting with the infants and their mothers following either frenotomy or sham procedure were unaware which group the infants were assigned to.

Following the initial procedure (frenotomy or sham), infants breastfed and researchers

assigned a LATCH score and assessed nipple pain. Then infants were separated from their mothers again and received either frenotomy or the sham procedure, whichever they hadn't previously received, and then LATCH and pain scores assessed again.

Dollberg et al. (2006) reported overall decrease in maternal nipple pain and improved LATCH scores at the completion of the study. Results reported compiled scores of both groups following frenotomy for all twenty-five mother/infant dyads. LATCH and standard visual analogue pain scale scores comparing results directly after the first frenotomy or sham procedures were not provided but are stated to be more positive within the frenotomy group. For all mother/infant dyads following frenotomy, mean pain scores decreased from 7.1 ($SD=1.9$) to 5.3 ($SD=2.2$) ($p = .001$), while mean LATCH scores improved from 6.4 ($SD=2.3$) to 6.8 ($SD=2.0$) ($p = .06$). There was a statistically significant difference in pain scores but not LATCH scores between the groups, although details on how the study was powered were not provided. Accurate comparisons between the two groups cannot be determined without full disclosure of results. This study did use a slightly more objective measure of breastfeeding through use of the LATCH score but was also highly dependent upon maternal report of pain.

A 2011 randomized control trial by Buryk et al., examined the effects of frenotomy on maternal nipple pain and breastfeeding adequacy for infants under one month of age with significant ankyloglossia. Ankyloglossia severity was determined using the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF), maternal nipple pain was assessed using the Short-Form McGill Pain Questionnaire (SF-MPQ), and breastfeeding adequacy was assessed using the Infant Breastfeeding Assessment Tool (IBFAT). Infants were randomly assigned to frenotomy ($n=30$) or control ($n=28$) and mothers were blinded. Directly after the procedure, outcomes were measured using the SF-MPQ and IBFAT. Follow up

occurred again at two weeks.

Nipple pain scores improved in both groups although there was a larger improvement within the group that had received frenotomies ($p < .001$). Sample size was determined by seeking to see a change in *SD* of the pain score by 1.5 with an effect size of 0.41, power of 80% and $p < .05$. Breastfeeding adequacy was improved only within the intervention group although with a small effect size ($p = .029$). By two weeks, the groups did not differ in nipple pain scores, although by that time only one control infant had yet to receive a frenotomy. Buryk et al. (2011) did show improved nipple pain immediately following frenotomies and recommended consideration of the procedure for selected infants.

Berry et al. (2012) sought to determine whether previously reported improvements in breastfeeding following frenotomy were due to placebo effect or not. Their study was unique in that it was double blinded, used a consistent observer, and attempted to be objective on measuring outcomes. Infant/mother dyads were randomized to receive frenotomy or no intervention. Mothers were asked their pain rating (1-10 scale) during a two-minute preintervention feed and an observer used a modified LATCH and Infant Breastfeeding Assessment Tool (IBFAT). Infants were separated from parents, and then returned either after frenotomy or sham procedure. Mothers were asked whether they felt there were changes in breastfeeding and to re-rate their pain. The observer again assessed feeds based off of the LATCH and IBFAT tools. Infants in the no-intervention group then had frenotomies performed after reassessment of the outcomes following the sham procedure. Mothers were also phoned one day and three months post intervention and asked about their breastfeeding journey. At the three

month follow up, all mothers stated they would choose to have the procedure done again if in the same situation. No mothers reported increased breastfeeding difficulties at three months.

Twenty-seven infants received frenotomies while thirty were randomized to the control group. Seventy-eight percent of mothers from the intervention group felt breastfeeding had improved compared to 47% of the control group. Additionally, 77% of mothers whose infants had frenotomies correctly identified which group their infant was in while only 55% of the control group mothers did. There was no statistically significant difference in reported nipple pain ($p = .13$) or observed breastfeeding changes between the groups (p value not provided).

Mothers did report immediate subjective improvements following frenotomies. Continued breastfeeding rates for all participants were roughly double the national average, although because the control group also received the intervention after having no procedure, true comparisons in breastfeeding rates between the control and intervention groups were not feasible. Berry et al. (2012) concluded that improvements seen were not solely a placebo effect, and that frenotomies are safe and acceptable by parents.

A study in 2014 by Emond et al. investigated whether frenotomy within the first two weeks of life for infants with mild to moderate ankyloglossia was superior to standard care of lactation support. The sample was selected from a lactation clinic. Inclusion criteria included mild to moderate ankyloglossia defined as a HATLFF score of 6-12, breastfeeding difficulties defined as a LATCH score of 8 or below in a full term infant with a less than 10% weight loss. Infants were randomized to receive frenotomy ($n=53$) or standard care ($n=52$), and breastfeeding measures were assessed again at five days and eight weeks by a blinded observer. The control group was then offered frenotomy.

The primary outcome was the assessed LATCH score. No difference in LATCH ($p = .52$) or IBFAT ($p = .36$) scores were observed between the groups. Additionally, no difference in nipple pain via the visual analogue scale was seen ($p = 0.09$). Both groups had higher breastfeeding self-efficacy assessed by the Breastfeeding Self-Efficacy score-short form (BSES-SF) although the results were statistically significant only at the five day mark. The control group had higher rates of bottle feeding at five days. While no objective improvements were seen, the authors report that frenotomy was associated with improved self-efficacy and fewer families switching to formula.

Recent Research

Martinelli et al. (2015) looked at outcomes not previously measured. They were interested in breastfeeding patterns including number of sucks and pause lengths between suck clusters pre/post frenotomy. The researchers developed their own protocol to assess for ankyloglossia which involved history, anatomical and functional evaluation and sucking evaluation. Yes/no questions about breastfeeding were asked to mothers ($n=109$) of infants that were thirty days old. Based on the authors' tool, fourteen infants were selected to receive frenotomies, and an additional fourteen were non-randomly selected to serve as a control group; they were selected for gender and birth order to match the intervention group. Frenotomies were done on day forty-five of life and outcomes measured on day seventy-five. The same yes/no questions about feeding were repeated. Presumably a feed was observed to calculate number of sucks and pauses, although it is slightly unclear. Infants who received frenotomies had an increased number of sucks and decreased pauses between suck clusters, while the control group remained unchanged. Average sucks for the intervention group was 19.36 and increased to 53.75 at seventy-five days of life (55.76 to 54.50 for control group). Pause length decreased from 6.14sec to 3.88 sec (3.00 to 3.30 for control group). The authors state that frenotomies are a safe

and effective intervention for ankyloglossia. This study was limited by a tiny sample size without blinding or random assignment. Additionally, there is no standardized norm for number of sucks within sucking clusters or length of pauses between suck clusters for breastfeeding.

A 2017 study sought to explore the effects of frenotomy and reasons parents pursued the procedure (Muldon et al., 2017). The study had a survey design; all infants presenting to seven clinics within Ireland for a frenotomy were given the survey at their appointment for baseline measures and asked to return the survey within one week. Those who did return the survey were mailed a second survey a month later to assess post frenotomy outcomes. The surveys were subjective, completed by mothers, and included a modified LATCH score and questions about feeding, nipple pain with VAS pain score, demographics and referral source. Two-hundred-eighty-one initial surveys were distributed, 98 of which were completed (35% response rate). Eighty-nine mothers completed the second survey, which was 91% of those who were mailed the second survey. Exclusive breastfeeding rates did not differ one month post frenotomy (58%), although rates of strictly formula feeding among infants who were previously breast and bottle fed doubled.

Private lactation consultants were the primary referral source and most parents reported poor latch followed by nipple pain as the main reasons to pursue a frenotomy. Overall, 91% of mothers reported improved breastfeeding, and VAS pain scores decreased from 5.6 to 2.7.

Generally speaking, outcomes were favorable although there may have been response bias with only 35% of sample completing the initial questionnaire.

In 2019, Schlatter et al. conducted a study on causes of breastfeeding issues with a focus on frenotomies. Infants of thirty-five weeks or greater gestation with breastfeeding issues who were born at one hospital within Germany during the study period were included. Of the 776 births, 345 had breastfeeding issues (44%), and of them 116 had ankyloglossia (15%). Initially,

mothers were given questionnaires about their perception of breastfeeding, including a ten point pain scale, LATCH scores, and the Bristol Breastfeeding Assessment Tool (BBAT). Thirty had frenotomies conducted.

Frenotomies were offered if infants had a HATLFF score below eleven. Two and half weeks post-frenotomy LATCH, BBAT, and pain scores were collected via telephone. Ankyloglossia, first time breast feeder, low birth weight, late preterm and birth by cesarean were all associated with breastfeeding issues. LATCH scores improved from 6.9 to 9.5 in the infants who received frenotomies and from 7.5 to 9.5 for those with ankyloglossia without frenotomies (Mann-Whitney *U* test showed significant improvement with $p = 0.044$). The authors did not report BBAT or breastfeeding pain measures so no other assessment items were available for comparison. Only 45% of infants with ankyloglossia had breastfeeding issues. This study was unique in that it included late preterm infants, and that frenotomies were offered in the first few days of life based on low HATLFF scores. Similar to other studies, the sample size was small, there was no blinding, and there was reliance on self-report measures.

Posterior Ankyloglossia and Labial Frenulums

More recent research on ankyloglossia has focused on the use of frenotomy for posterior ankyloglossia and labial frenulums. Posterior ankyloglossia continues to be a controversial topic without an agreed upon definition. In general, there is thought to be posterior tongue-tie when the lingual frenulum inserts towards the posterior aspect of the tongue, or in some cases with no visual frenulums but when a fibrous band of tissue can be palpated within the submucosa underneath the tongue. Labial frenulums are the presence of a fibrous tissue between the gums and the lip. Labial frenulums are thought to inhibit flanging of the lips during breastfeeding, thereby impacting latch.

Benoiton et al. (2016) examined the effects of frenotomy for posterior ankyloglossia and upper labial frenulums. Infants with feeding difficulties and either posterior ankyloglossia or

labial frenulums were referred from lactation consultants to one otolaryngology clinic. The HATLFF tool was used to assess frenulum function although no cutoff scores were reported.

Additionally, the HATLFF tool does not distinguish between anterior and posterior ankyloglossia, therefore the inclusion criteria for posterior ankyloglossia was subjective.

Breastfed and bottle-fed infants were included. Frenotomies were performed and then either a breastfeeding session was observed or if the infant refused to nurse, a follow up phone call was conducted within twenty-four hours and at two weeks. Outcomes were mothers' subjective report of latching issues, painful nipples, poor weight gain, clicking noises or unsettled feeds.

Frenotomies were conducted on thirty-four babies. Twenty-one had lingual frenotomies only, ten had lingual and labial and only three had labial frenotomies solely. Eighty-five percent of infants had immediate improvements in breastfeeding and 82% had continued improvements at the two week follow up. Forty-one percent of the babies had already had a previous frenotomy procedure for anterior ankyloglossia and an additional two infants had revision procedures following the study. Rates of breastfeeding vs. bottle feeding were not reported, nor were outcomes measured objectively. Comparing outcomes amongst the groups was impractical due to small sample size.

Ghaheri et al. (2017) examined the effectiveness of lingual and labial frenotomies on nipple pain, infant reflux and breastfeeding self-efficacy. While their study did not seek to specifically study posterior ankyloglossia, 78% of the infants involved had isolated posterior ankyloglossia defined as a Type III or IV category on the Coryllos tongue-tie classification system. Labial frenulums were assessed with the Kotlow upper lip-tie classification system. Measures were taken at baseline, one week and one month post procedure through an electronic survey. The revised infant gastroesophageal reflux questionnaire (I-GERQ-R), BSES-SF and

VAS for nipple pain were used at each point of follow up. Breastmilk intake at baseline and one week were also measured via pre/post feed weights for sixty infants. Infants were all referred to the principal investigator for frenotomies. They had to be less than twelve weeks old and having breastfeeding issues; 237 infants had frenotomies [75% (n=178) labial and lingual, 0.42% (n=1) labial only, 24% (n=58) lingual only].

Between each data collection point, improvements were seen with each outcome. Self-efficacy was measured using the BSES-SF. At baseline the BSES-SF mean score was 43.9, which increased to 52.3 at one week and 56.5 at one month ($p < .001$). Infant reflux was assessed using the I-GERQ-R, with lower scores being associated with decreased reflux symptoms. I-GERQ-R scores went from 16.5 at baseline to 13.2 and 11.6 at one week and one month follow up ($p < .001$). VAS for pain also decreased from 4.6 initially to 2.2 and then 1.5 at the one month mark ($p < .001$). Authors reported improved breastmilk intake of 155%, from 3ml to 4.9ml per min of breastfeeding, measured in sixty infants with pre/post feed weights ($p < .001$). The study was powered to detect a two-point change in BSES-SF, which has commonly been observed in control groups in other studies on ankyloglossia. This study was the first to look at reflux; the authors conducted a subsequent study also looking at reflux and ankyloglossia. Most infants received both labial and lingual frenotomies, comparisons between groups were not feasible due to the low number of infants receiving only one type of procedure. The outcomes were all subjective measures, and the study had no comparison group or blinding.

Ideal Timing of Frenotomies

The majority of studies on ankyloglossia have involved infants within the first three months of age. Frenotomies have been performed on infants to facilitate breastfeeding from several days old as in the Schlatter et al. (2019) study up to nine months of age such as in the Ghaheri et al. (2018) study. There have been two studies to date addressing optimal timing of frenotomies.

The first study to look at ideal timing was a retrospective survey conducted in 2012 by Steehler et al. They performed a chart review to identify all infants who had been seen in one clinic for ankyloglossia and then conducted telephone surveys asking about how helpful parents thought frenotomies were and how long they breastfed for. Three-hundred-sixty-seven infants had been seen for ankyloglossia, 301 infants had received a frenotomy, 91 parents participated in the survey (25% response rate, 82 of whom had had frenotomies while 9 had not). Eighty-three percent of infants who had had frenotomies continued breastfeeding while only 67% of those without frenotomies had, although both groups nursed for an average of 5.8 months. Out of the infants who had the procedure during their first week of life, 86% of mothers felt it significantly improved breastfeeding while only 74% of mothers of babies with frenotomies after the first week of life felt it significantly improved breastfeeding.

Sharma and Jayara (2015) conducted a survey of all infants who had been diagnosed with ankyloglossia within a twelve month period at one clinic. Fifty-four infants were included, and 81% (n=36) had frenotomies; 78% of those completed the survey. Telephone surveys of the IBFAT were completed prior to the procedure and one month following. Infants ranged in age from 15-178 days, with an average of 38 days. Eighty-one percent reported improvement with the average IBFAT scores increasing from 3.33 to 9.19. In infants less than thirty days old, 94% (n=17) improved compared to only 68% of those older than thirty days (n=19).

Frenotomy Revision

Several studies previously discussed have included infants who had already had a frenotomy completed before the initiation of the study. This is a confounding variable that is not usually accounted for. Ghaheri et al. (2018) sought to determine the effects of lingual and labial frenotomy revision for infants with continued feeding problems on breastfeeding. The Coryllos

tongue-tie classification system was used to measure ankyloglossia, where Type I and II are classified as anterior and Types III and IV as posterior. The majority of the sample had posterior tongue-ties (83%). The Kotlow upper lip-tie classification system was used to objectively assess labial frenulums. Fifty-four infants who were all referred to one clinic for ongoing feeding issues had repeat frenotomies (37% lingual only, 63% lingual and labial). Follow up was completed at one week and one-month post procedure and included the BSES-SF, VAS for nipple pain and the I-GERQ-R.

Improvements were seen in all three outcomes measured at both follow up points. At one week post procedure, 18% had improved BSES-SF and I-GERQ-R scores and 50% had improved VAS nipple pain scores. Improvements continued at the one month follow up but less overall change was observed. For example, the mean VAS pain score at baseline was 4.8, 2.2 at one week and 1.6 at one month. I-GERQ-R scores were 15.7 at baseline, 11.9 at one week and 10.4 at one month post procedure, with lower scores being associated with decreased reflux symptoms. The BSES-SF scores increased over time with a mean of 45.1 at baseline, to 52.1 at one week and 56.9 at one month. Scores above fifty are associated with higher self-efficacy and continued breastfeeding. Sample size was determined assuming 80% power, $p < .05$, $r = .5$, to detect a 5-point change in the BSES-SF tool. All three outcomes had statistically significant improvements; BSES-SF ($F = 41.2$, $p < .001$), I-GERQ-R ($F = 22.7$, $p < .001$), and VAS pain scale ($F = 46.1$, $p < .001$).

Laser vs. Scissors

The Ghaheri et al. (2017) and Ghaheri et al. (2018) studies on ankyloglossia both used light amplification by stimulated emission of radiation (LASER) to perform frenotomies. These two studies used similar outcome measures as other studies on ankyloglossia but direct

comparison of outcomes are somewhat limited due to methodological shortcomings. The rest of the studies in this review used sterile scissors to perform frenotomies. There have been no direct studies comparing frenotomies with sterile scissors vs. laser for infants.

A review of the use of lasers for ankyloglossia for children under fifteen years of age found lasers to be preferable to standard treatment (Garrocho-Rangel et al., 2019). Standard treatment was described as the use of a scalpel, although studies that describe frenotomy procedures typically describe the use of scissors. The review included five case studies, eight narrative reviews, three cohort studies and one RCT. The only RCT involved ten teenage patients. Garrocho-Rangel et al. (2019) state laser is superior due to decreased surgical time, no need for sutures, less requirement of anesthetic and pain medication post procedure, and decreased bleeding. The authors' conclusions are not applicable to the neonatal population. Suturing for frenotomies is usually only done for procedures under general anesthesia. Typically, there is no anesthetic used in neonates, no pain medications given post frenotomy, minimal bleeding is reported across studies, and the procedure is very quickly done with scissors and suturing is not required.

Provider Perception

With limited high-quality studies on ankyloglossia, and no standard objective definitions or guidelines, it is not surprising that opinions about ankyloglossia vary widely throughout the medical community. There have been two surveys conducted which have looked at provider perception.

The first survey was conducted in 2000 by Messner and Lalakea. They surveyed 423 otolaryngologists, 425 pediatricians, 400 speech pathologists, and 350 lactation consultants. The sample was randomly selected from professional membership organizations. Response rates

were 49%, 55%, 38% and 58% respectively. All groups reported that they infrequently encountered ankyloglossia. Lactation consultants were most likely (69%) to feel ankyloglossia is frequently associated with breastfeeding issues. Conversely, 90% of pediatricians and 70% of otolaryngologists felt ankyloglossia is never or rarely associated with breastfeeding difficulties.

The second survey included 400 Australian surgeons (Brinkmann et al., 2004). Authors did not report how respondents were selected. A questionnaire was developed using the Dillman's Total Design Method and had questions pertaining to referrals, assessment, surgical procedure technique, indications for frenotomies, and outcomes. The overall response rate was 81%, with 46% being oral/maxillofacial surgeons, 37% plastic surgeons and 17% general pediatric surgeons. Overall, 10% believed that frenotomies were rarely needed; this thought was prevalent in 17% of general pediatric surgeons. The majority thought decreased tongue motility was an indication for frenotomy. While most thought that frenotomies were successful, 1.3% thought they were successful less than 25% of the time. There were no universally agreed upon ways to assess for ankyloglossia, specific indications for frenotomies, or consistent expected outcomes.

Community Efforts to Decrease Frenotomies

Providers of newborn care in Canterbury, New Zealand were observing great increases in frenotomy procedures. In an effort to avoid unnecessary frenotomies while also trying to identify newborns that would most likely benefit from frenotomies, a region-wide program was developed to standardize assessment and recommendations pertaining to ankyloglossia (Dixon et al., 2018). An interdisciplinary team involving all stakeholders developed and agreed upon a consensus pathway using the BTAT tool. Initially, infants with a BTAT score of five or less, which was later decreased to four, were offered frenotomies with the ideal age of the procedure

being 2-4 weeks. Following either lactation support for infants with higher BTAT scores, or frenotomy procedures when indicated, families were telephoned 31-193 days later and asked about feeding method, difficulties and perception of feedings. Additionally, websites with educational materials aimed for both families and healthcare providers were developed to help create buy-in and support. A total of 309 infants were found to have ankyloglossia, and 264 had frenotomies. Throughout the course of the project, frenotomy rates in Canterbury decreased from 11.3% to 3.5% while feeding methods of infants who both received and did not receive frenotomies did not change. A total of 177 infants had follow up (62% of frenotomy group, 29% of non-frenotomy group). Eighty-five percent of mothers of infants who had received frenotomies reported improved breastfeeding, while 49% of the non-frenotomy group reported no lasting breastfeeding issues. Rates of exclusive breastfeeding following frenotomies were greater for those with BTAT scores of four or less compared to five (38% to 54% compared to 48% to 53%). Feeding methods between infants who had and had not received frenotomies did not vary pre/post intervention.

The low response rates were the main limitation for assessing outcomes. The consensus pathway development and implementation was significant in that it involved a community tackling what they believed to be a large issue; they were able to standardize their approach, reducing frenotomy rates while not negatively impacting breastfeeding rates.

Discussion

The majority of studies on ankyloglossia have been limited by small sample size, no comparison group, no blinding to the intervention, limited follow up with no long-term outcomes, lack of true control groups, and relying upon subjective data including pain scores, self-report of breastfeeding issues, subjective assessment of ankyloglossia and breastfeeding self-

efficacy. Of the five RCTs, one of the studies had no blinding, two were double blinded while the other two were single blinded. Some of the researchers attempted to be objective through the use of many tools to assess breastfeeding adequacy, ankyloglossia, breastfeeding self-efficacy, infant reflux symptoms and maternal nipple pain, although the tools used still highly depend upon subjective measures. Participants had either no follow-up or follow-up ranging two to twelve months, although conclusions about long term effects of frenotomies could not be drawn because the majority of the control groups in all the RCT studies elected to eventually have frenotomies done.

There has been much controversy over whether frenotomies are appropriate for ankyloglossia and whether they actually positively impact breastfeeding. Overall the studies did show improvements in subjective measures of breastfeeding, but failed to show improvements in objective measures such as LATCH score. Hogan et al.'s (2005) study showed the most beneficial results with an overall improvement in infant feeding by 97% following frenotomy, although the study lacked rigor in its design, and likely had a large placebo effect. Dollberg et al.'s (2006) study showed improvement in nipple pain and LATCH score but failed to truly compare the control and intervention groups. Buryk et al.'s (2011) study showed a placebo effect with the control group also reporting improved nipple pain although the intervention group had greater improvements. Forty-seven percent of the control group in Berry et al.'s (2012) study reported improved breastfeeding despite not receiving frenotomies, but this was lower than the 78% self-report of improvement in the intervention group. While there could be placebo effect, there also does appear to be a real change in subjective reports of breastfeeding. Nipple pain outcomes were inconsistent between the studies. Pre/post intervention assessment of breastfeeding through LATCH and/or IBFAT scores did not differ statistically in Buryk et al.'s

(2011), Berry et al.'s (2012), or Emond et al.'s (2014) studies.

Breastfeeding is dynamic and complex and can be influenced by many factors. The discrepancy in improved results with subjective vs objective measures of breastfeeding calls into question the validity of the assessment tools used. Are the tools adequate to assess changes in breastfeeding that result from frenotomies? There could be very real improvements that the tools are not sensitive or specific enough to detect. Many of the tools used also relied heavily upon subjective report.

Frenotomies were shown to be safe and acceptable to parents. No significant complications were reported. The majority of control group mother/infant dyads elected to have frenotomies completed. In Hogan et al.'s (2005) study one infant in the control group improved with lactation support alone, the remaining twenty-eight elected to have frenotomies done after forty-eight hours. All mothers from Berry et al.'s study (2012) stated they would choose to go through with a frenotomy if in the same situation again. All mothers who met inclusion criteria in the Buryk et al. (2011) study participated. Only one study reviewed reported any negative effects of frenotomy. In O'Callahan et al.'s 2013 study, 6% of parents reported negative emotional or physical effects of frenotomy. While the negative effects were not explicitly stated, it may have been related to the frenotomy technique used. This study focused on posterior ankyloglossia, graphically described dissection of the frenulum, and the author mentioned all eight providers he had trained to perform posterior frenotomies were uncomfortable doing so. The negative effects may have been more related to the researchers' aggressive management as opposed to the more customary care.

Among the three studies that reported national averages for breastfeeding, infants who received frenotomies had either higher breastfeeding rates than the national averages or were less

likely to be formula feeding post intervention (Berry et al., 2012; Buryk et al., 2011; Emond et al., 2014). While the participants were likely highly motivated to succeed at breastfeeding, this does support the thesis that frenotomies do help improve breastfeeding. In contrast, the Dixon et al. (2018) article found no change in infant feeding method following decreased frenotomies within Canterbury, NZ, although results were not based on national or regional reports but rather telephone follow up. This was a unique approach to the management of ankyloglossia which has not been described elsewhere.

There remain many unanswered questions about ankyloglossia. The identification of posterior tongue-tie is controversial at best. While the Kotlow and Coryllos classification systems identify anterior vs posterior ankyloglossia, the literature has not identified whether anterior or posterior ankyloglossia has a greater impact upon infant feeding. Studies addressing posterior ankyloglossia were plagued by small sample sizes and often infants that also had labial frenotomies simultaneously, limiting ability to generalize outcomes. There remains no link between severity of ankyloglossia and feeding issues. The HATLFF and BTAT tools do address functional aspects of the tongue and frenulum, making them superior to the Kotlow and Coryllos classification systems. While the HATLFF is burdensome and challenging to use, the BTAT is much simpler and straight forward and therefore could become the gold standard for assessing ankyloglossia moving forward. Only two small studies have directly addressed timing of frenotomies. They both suggest earlier frenotomies to be superior. It is unknown whether this finding is a result of improvements that occur naturally in the early stages of mother/infant dyads becoming accustomed to breastfeeding or truly indicative of the superiority of early frenotomy. No studies have directly compared frenotomy procedures (laser vs scissors, etc.) to determine if one method is more ideal. Numerous studies involved infants who had already had a frenotomy,

and some went on to have revisions. One study directly addressed revisions and found them to be beneficial. If frenotomies have such positive outcomes, it is unknown why revisions would be necessary.

Missing from the literature are consequences of the natural history of not performing frenotomies. Some authors have also promoted frenotomies to prevent social issues such as the inability to lick ice cream, kiss or play specific instruments and the possible effects on self-esteem or other psychological issues (Chinnadurai et al., 2015). There does not appear to be a plethora of adults today who feel negatively impacted by not having had their frenulums surgically removed as infants as would be expected if there was significant psychological sequelae of not intervening. Often benefits of intervening appear to be overemphasized within the literature reflecting the clear biases of authors.

Frenotomies for the management of ankyloglossia do consistently show decreased maternal nipple pain. Other outcomes such as infant latch, breastfeeding quality and self-efficacy, are less clear. Overall, frenotomies are a low risk procedure and selected mother/infant dyads are likely to benefit from them. Major limitations to studies on ankyloglossia include relatively small sample sizes, limited blinding, inability to compare groups long term because the majority of the control groups received the intervention, and potentially ineffective measurement tools. Much is still unknown about ankyloglossia. Further research is needed to determine optimal timing of frenotomies, surgical technique, development of tools that can better measure breastfeeding outcomes specifically related to frenotomies, studies with larger sample sizes for increased power, and examination of long-term outcomes.

Theoretical Model and Methodology

Theoretical Model

Havelock's Theory of Change is based on Lewin's Planned Change Theory (Havelock, 1973). It encompasses six steps for planned change: building a relationship, diagnosing the problem, gathering resources, choosing the solution, gaining acceptance and stabilization and self-renewal. It was initially developed for use within education although it has been applied to nursing as well. Havelock's Theory of Change focuses on the incorporation of knowledge into practice (Havelock, 1973).

Havelock's Theory of Change is well suited for this quality improvement study. Initially while building rapport with colleagues, discussions on the need for change in the management of ankyloglossia will begin. In the second step, the need for change of the management of ankyloglossia will be reaffirmed. Next, the systematic review of the literature will show what is known and what is still lacking in knowledge pertaining to ankyloglossia in the newborn and help to direct further steps. Finally, a new clinical approach to management of ankyloglossia will be selected, incorporated into practice, monitored, and hopefully will become the new norm.

Project and Study Design

This is a quality improvement project that will focus on standardizing the assessment and management of ankyloglossia and exploring providers views on ankyloglossia within a private pediatric practice in Albuquerque, New Mexico that has approximately 30 providers.

An educational in-service will be conducted addressing what is currently known about ankyloglossia. At this in-service, providers will be trained on using the Bristol Tongue Assessment Tool (BTAT). This tool was selected due to its ease of use and ability to also assess functional impairment. Infants with a BTAT score of four or less (0-3 is considered severely

impaired) with breastfeeding difficulties would be offered frenotomies. This threshold is based on Dixon et al.'s 2018 study where a final cut off of four was also selected and no change in feeding patterns were seen between infants who had or had not received frenotomies.

Varying perceptions about ankyloglossia have been reported amongst healthcare provider groups with pediatricians in general believing there is less of an association between ankyloglossia and breastfeeding difficulties. This study will also look at the perceptions of healthcare providers and nurses within a private pediatric setting. A questionnaire will be developed and administered prior to the educational in-service. Questions will focus on ankyloglossia, its relationship to breastfeeding, indications for frenotomies, experience with frenotomies, assessment of ankyloglossia and expected outcomes following frenotomy. An immediate post-test questionnaire and one six weeks after the educational in-service will be conducted, focusing on whether providers have incorporated the tool into their clinical practice, whether they feel the tool is useful and practical, and whether or not the perception of ankyloglossia has changed following the educational in-service.

Setting and Resources

This study will take place at After Hours Pediatrics. After Hours Pediatrics is the largest pediatric private practice within New Mexico. There are a total of four ambulatory care clinics, three located in Albuquerque and one in Rio Rancho. Many of the staff, providers included, float between clinics. The clinics are all under the same management, and share the same electronic medical records system. Resources needed for the project include a computer and projector to use for the educational in-service, printed handouts reflecting the in-service content, and individual use of computers to complete online questionnaires.

Study Population

The study population includes all healthcare providers and nurses who provide newborn care at any of the After Hours Pediatrics Clinics. Currently, three of the clinics have a nurse; one is a licensed practical nurse and the other two are registered nurses. These nurses conduct weight checks for newborns whose weight is borderline or have had excessive weight loss. They are often problem solving feeding difficulties with families. The nurses are included in this study because they regularly interact with newborns with feeding difficulties, and may identify infants with ankyloglossia and feeding challenges more regularly than some of the providers. The providers include all pediatricians, pediatric nurse practitioners, family nurse practitioners and physician assistants employed at After Hours Pediatrics. Currently there are a total of twenty-nine providers.

Providers or nurses were be excluded if they never provide newborn care. Educational in-services were scheduled at each clinic during the lunch-break hour to facilitate participation. Emails were sent out to all providers and nurses for recruitment, explaining the project and asking for them to sign up for one of the educational in-services. Providers and nurses that did not sign up for the educational in-service were followed up via email and/or personal contact to encourage participation.

Sources of Data

The primary source of data for analysis was the three questionnaires administered prior to, immediately after, and six weeks following the educational in-service. The questionnaires were completed by the providers and nurses who have attended the in-services. The first questionnaire focused on ankyloglossia, its relationship to breastfeeding, indications for frenotomies, experience with frenotomies, assessment of ankyloglossia, and expected

outcomes of frenotomies. The follow up questionnaires focused on whether providers had incorporated the BTAT tool into their clinical practice, whether they felt the tool was useful and practical, and contemporary perceptions of ankyloglossia to ascertain any changes following the educational in- service.

Data Analysis

The initial two questionnaires were done via paper at the start and completion of the educational in-service. They were placed in two separate folders. The final questionnaire was conducted via REDCap. Questions pertaining to perception of ankyloglossia's relationship to breastfeeding were repeated on the follow up questionnaires. Questions focused on preexisting knowledge about ankyloglossia, frequency of encountering ankyloglossia in clinical practice, knowledge of frenotomies, and any changes in perception of ankyloglossia or clinical practice six weeks following the educational in-service. Data were analyzed to ascertain whether views on ankyloglossia have changed following the educational in-service.

Quality

The third questionnaire was conducted via REDCap which allows for responses to be confidential (Patridge & Bardyn, 2018). The first two were hand written and in person to encourage completion. They were placed by participants into folders to maintain confidentiality. As a quality improvement project, the sample size is somewhat limited to only the providers and nurses working at After Hours Pediatrics. To ensure optimal participation, multiple educational in-services were held during the lunch hour and at each location, and the population was personally contacted when possible to encourage participation. There was opportunity for participants to ask questions about the BTAT tool and ankyloglossia in general at the educational in-service and afterwards either via email or in person.

Ethics and Human Subjects Protection

This project was submitted for approval by the University of New Mexico's Institutional Review Board. There are no ethical issues with this study. The study participants were healthcare providers and nurses. There were no vulnerable population groups. All participants had the right to withdraw from the project at any point. There were no serious risks as this was an educational in-service with the goal of creating a standardized approach to managing ankyloglossia. Benefits for providers included increased knowledge of ankyloglossia and learning about an objective tool to better assess tongue-tie. Potential benefits for the community include avoiding unnecessary frenotomies while recommending them for the infants most likely to benefit from the procedure.

Timeframes or Timeline

The project was submitted to the IRB in December 2019 and approved in January of 2020. The educational in-services occurred over the first two weeks of February of 2020 and the six week follow up survey conducted electronically six weeks later in March. The pretest and posttest were completed at the start and completion of the educational in-services via pen and paper. The follow up questionnaire was completed via REDCap through workplace emails. Data was analyzed late Spring and early summer of 2020.

Budget

At the completion of the six-week questionnaire, participants received a five-dollar merchandise card to Starbucks. This was the only monetary item involved in participation. The clinic has a projector that was used for the educational in-service, and all employees have access to computers which could have been used to complete the questionnaires. To be respectful of people's clinic time, the in-services occurred during the lunch break.

Strengths and Weaknesses of the Study

The main strength of this project has been to establish an evidence based standardized approach to the assessment and management of ankyloglossia in this clinic setting. This allowed providers to present a united front and decrease parental confusion from numerous conflicting recommendations that are often received. Additionally, reassurance can be provided to parents whose infants have adequate frenulum function based on the assessment of the BTAT. Overall, this should allow for infants who may most benefit from a frenotomy to receive that recommendation while avoiding unnecessary procedures for those less likely to benefit.

There are several limitations to the impact of this project. While the BTAT tool is more user-friendly than its predecessors, there is still no agreed upon criteria for diagnosis of ankyloglossia or set criteria for when frenotomies should be done. The parameters selected for this study have been based on the previous limited research. Additionally, pediatricians have traditionally been skeptical of the relationship between ankyloglossia and breastfeeding difficulties. This had the potential of creating challenges when asking all providers to start incorporating the use of the BTAT tools and assessing frenulums within their clinical practice. The study timeframe was also somewhat short out of necessity. Ideally the use of the BTAT will continue well after the follow up period, allowing for a continued standardized approach to the assessment and management of ankyloglossia.

Results and Discussion

Results/outcomes

Twenty providers and nurses attended the educational in-services on ankyloglossia. Due to staffing changes between the project proposal and educational in-services, there was a total of forty-one eligible people invited to participate, therefore 49% of eligible people participated. Of them, eleven were pediatricians, two were physician assistants, four were nurse practitioners and

three were nurses. All twenty completed the initial questionnaire or the pretest. Eighteen completed the posttest, which was the second questionnaire immediately following the educational in-service. Of the two participants that did not complete the posttest, one needed to attend to a sick patient present during the lunch break and the second left the in-service during the questions portion at the conclusion of the in-service. Shortly thereafter they were provided with the posttest, but it was never returned. Therefore, total response rate for the posttest was 90%. One provider left the practice before the six week follow-up test. Seventeen participants completed the six week follow-up test resulting in an 85% response rate for the follow-up test.

Several questions were repeated on multiple tests to assess whether the educational in-service changed provider perception on tongue-tie and related topics. Tables 1-4 show the questions that were asked more than once. Table 1 focuses on perception of tongue-tie prevalence. Perception of prevalence initially was more spread out although after the in-service 67% reported prevalence of tongue-tie as 5-10%.

Table 1

What percentage of newborns have tongue-tie? (frequency/percentage)

	0-5%	5-10%	10-15%	15-20%
pretest	7(35%)	7(35%)	5(25%)	1(5%)
posttest	3(20%)	10(67%)	2(13%)	0(0)%

Table 2 shows results for perception of the impact of tongue-tie on breastfeeding. Initially, 50% of respondents answered that tongue-tie occasionally affected breastfeeding, 20% answered sometimes, while only 5% of respondents answered most of the time. In the second questionnaire, a larger percentage (39%) of respondents answered sometimes, although the same number of respondents remained undecided (2) or reported that it hardly ever has an effect (3). In the follow-up test, the responses were most similar to the pretest with 59% answering

occasionally and 29% answering sometimes. Interestingly, zero respondents marked undecided, and zero responded never or always for all three tests.

Table 2

How often does tongue-tie negatively impact breastfeeding? (frequency/percentage)

	Never	Hardly ever	Occasionally	Undecided	Sometimes	Most of the time	always
pretest	0(0%)	3(15%)	10(50%)	2(10%)	4(20%)	1(5%)	0(0%)
posttest	0(0%)	3(17%)	6(33%)	2(11%)	7(39%)	0(0%)	0(0%)
Follow-up test	0(0%)	2(12%)	10(59%)	0(0%)	5(29%)	0(0%)	0(0%)

The percentage of providers who thought frenotomies were beneficial for breastfeeding decreased following the educational in-service. Table 3 shows respondents' answers. Initially, 60% agreed (slightly agree/agree), 25% were undecided, and 15% disagreed (disagree/slightly/disagree). After the in-service only 34% agreed (slightly agree/agree) while 50% were undecided and 17% disagreed (disagree/slightly disagree). Table 4 shows perceptions about the side effects of frenotomies. Initially all respondents thought frenotomies were well tolerated with few side effects. In the posttest one person disagreed and two were undecided.

Table 3

Do you agree with the following statement: frenotomies help improve breastfeeding for infants with tongue-tie? (frequency/percentage)

	Strongly disagree	Disagree	Slightly disagree	Neither agree nor disagree	Slightly agree	Agree	Strongly agree
pretest	0(0%)	1(5%)	2(10%)	5(25%)	9(45%)	3(15%)	0(0%)
posttest	0(0%)	2(11%)	1(6%)	9(50%)	5(28%)	1(6%)	0(0%)

Table 4

Do you agree with the following statement: frenotomies are generally well tolerated with few side effects. (frequency/percentage)

	Strongly disagree	Disagree	Slightly disagree	Neither agree nor disagree	Slightly agree	Agree	Strongly agree
pretest	0(0%)	0(0%)	0(0%)	0(0%)	4(25%)	11(69%)	1(6%)
Posttest	0(0%)	0(0%)	1(6%)	2(11%)	2(11%)	11(61%)	2(11%)

The pretest asked the open-ended question: “how do you assess tongue-tie?” Seventeen different responses were provided. Nine people indicated tongue protrusion as important, four mentioned assessing the frenulum (thickness/length/tight/short/attachment to tongue), three mentioned tongue shape (especially observing for heart shaped tongue), three referenced nipple pain, and two mentioned latch or breastfeeding issues. Other responses were more vague such as “clinically”, “inspect and feel”, “PRN, when parents ask”, and “[by]physical exam”. The pretest and posttest asked about indications, and expected outcomes for frenotomies; results are reported in Table 5. Indications for frenotomies were similar to assessment answers, and included pain/trouble breastfeeding, weight loss/suboptimal gaining, presence of tongue-tie (especially if tight), poor breastfeeding or latch, and speech/articulation issues. In the posttest four people discussed BTAT scores as an indication for frenotomy, additional indications included parental desire and two who were unsure. Expected outcomes of frenotomies at the pretest included improved tongue mobility/protrusion, latch, breastfeeding, weight gain, nipple pain, and articulation. Additional outcomes given in the posttest included parental satisfaction and parental perception.

Table 5*Indications and Outcomes of Frenotomies*

	pretest	posttest
What are indications for frenotomies?	<p><u>Feeding related</u></p> <ul style="list-style-type: none"> • Pain/trouble feeding, wt loss, not gaining • Trouble with oral/motor skills • Poor feeding/weight gain • Trouble feeding • Painful nursing and inadequate intake • Poor weight gain • Pain in mother with breastfeeding • Negative latching • Feeding problems, maternal nipple pain • Trouble nursing, gaining weight, nipple pain • Poor feeding • Failure to breastfeed • Poor feeding, poor weight gain, nipple pain • Difficulty latching or feeding • Difficulty feeding • Nursing difficulties • Nipple pain with breastfeeding, poor latch <p><u>Tongue anatomy</u></p> <ul style="list-style-type: none"> • Obvious ankyloglossia • Heart shaped tongue • Tight tongue tie Unable to stick tongue out beyond lip • Inability to protrude [tongue] for feeding <p><u>Speech related</u></p> <ul style="list-style-type: none"> • Speech concerns • Speech delay • Articulation errors • Articulation errors when older • Older child articulation • Speech problems 	<p><u>Feeding related</u></p> <ul style="list-style-type: none"> • Poor weight gain • Nipple pain, poor latch • Persistent nipple pain/feeding difficulty • Nipple pain after a few weeks of breastfeeding • Nipple pain, latching problems • Feeding issues • Trouble breastfeeding (latch, wt gain), nipple pain • Difficulty nursing, nipple pain • Trouble with nursing, maternal pain • Poor breastfeeding • Difficulty nursing, nipple pain • Pain, low weight gain <p><u>Tongue anatomy</u></p> <ul style="list-style-type: none"> • Tongue movement and structure <p><u>BTAT</u></p> <ul style="list-style-type: none"> • Persistent nipple pain and a BTAT <4 • Difficulty with breastfeeding plus evidence that tongue tie is present based on scoring system • Bristol scoring four or less in the scenario of troubled nursing • BTAT less than 4 and impaired tongue mobility <p><u>Other</u></p> <ul style="list-style-type: none"> • Parental desire • I'm not sure • Not known

<p>What are the main outcomes for frenotomies?</p>	<p><u>Feeding related</u></p> <ul style="list-style-type: none"> • Better latch • Improved feeding • Better feeding/growth • Less painful breastfeeding, more successful breastfeeding • Better latching • Improved feeding and nipple pain • Maternal report of improved feeding • Improved feeding • Better feeding, better suck quality • Improved latch, decreased nipple pain • Improvement in nursing <p><u>Speech related</u></p> <ul style="list-style-type: none"> • Improved speech • Improved articulation <p><u>Tongue anatomy/mobility</u></p> <ul style="list-style-type: none"> • Able to thrust tongue • Able to protrude tongue better and reach top of palate <p><u>Other</u></p> <ul style="list-style-type: none"> • 50/50 improvement • They always work, but I think a lot of it is maternal attitude • Sometimes helps, sometimes not • Not sure • Sometimes helps some • Small bleeding, no change 	<p><u>Feeding related</u></p> <ul style="list-style-type: none"> • Decreased maternal pain • Improved nipple pain • Improved pain • Reducing nipple pain, improve weight gain • Improved nipple pain and latching problems • Decreased nipple pain and better perception of breastfeeding • Improved breastfeeding • Improved nursing • Improvement in latch • Improved nursing and decreased maternal pain • Less breast pain • To help with breastfeeding • Better feeding, less pain <p><u>Other</u></p> <ul style="list-style-type: none"> • Parental satisfaction, questionable if necessary • Most improved per studies • Some mothers claim they help, other say no difference • Expensive • Not known • Research still undecided but parents are happy with procedure • Small bleeding, no change • Probably self fulfilling expectations • Improved parent perception
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Additional questions in the pretest asked about current practices related to tongue-tie. Forty-five percent of respondents regularly assessed for tongue-tie at well newborn visits (either slightly agree, or strongly agree). Ten percent neither agreed nor disagreed about regularly assessing for tongue-tie while 20% disagreed (strongly disagree/disagree). When asked if they encounter it in their clinical practice, similar percentages of providers reported encountering tongue-tie regularly. Fifty percent (slightly agree/agree/strongly agree) encountered tongue-tie regularly, 5% responded neither agree nor disagree, and 45% disagreed (strongly disagree/disagree/slightly disagree). When asked how often do parents of newborns ask you about tongue-tie, 0% reported never, 15% hardly ever, 20% occasionally, 0% undecided, 65%

sometimes, 0% most of the time and 0% always. Sixty-five percent of respondents had referred newborns to dentists or ENTs for frenotomies while only 13% (2 people) had performed frenotomies themselves. When asked if trained to perform frenotomies, would you be willing to perform them, 28% disagreed (strongly disagree/disagree), 7% (one person) neither agreed nor disagreed, one wrote in not applicable and 57% agreed (slightly agree/agree/strongly agree). Two people answered the last question of the pretest which asked about any additional thoughts about tongue-tie. One person wrote “I think the pediatric dental community is monetizing this condition”, while the other person wrote “There seems to be an increase in frenotomies in the last five years of my practice”.

The posttest asked participants to agree or disagree with the following statement: The BTAT tool is easy to use. While 6% (1 person) disagreed, 11% neither agreed nor disagreed, the majority at 84% agreed (slightly agree/agree/strongly agree). The posttest was administered directly following the educational in-service which focused on a review of the literature on tongue-tie and teaching the BTAT tool. The last question asked about additional thoughts; five people made comments, most of which focused on what is currently known on the topic, personal views and thoughts for future studies. Responses included “I would like to be better at assessing tongue tie and advising parents on the procedure”; “more evidence-based research needs to be performed”; “we need more research”; “for the most part not needed”; and “I feel every lactation consultants mentions tongue tie to parents. I’d like to have a survey of lactation consultants to see what percentage of babies are diagnosed with tongue tie”.

The follow-up test was completed electronically via REDcap six weeks following the educational in-service to allow time for providers to use the BTAT within their clinical practice. Only 18% of respondents reported using the BTAT with infants with tongue-tie and only 35% (6 people) had encountered tongue-tie within the follow up period. People were asked again to

agree or disagree with the statement: the BTAT is easy to use. This time 12% disagreed (disagree/slightly disagree), 12% neither agreed nor disagreed and 77% agreed (slightly agree/agree/strongly agree). Despite only 35% encountering tongue-tie, 53% reported that the BTAT had helped facilitate conversations with parents about tongue tie and 47% found the BTAT helped with making clinical decisions about management of tongue-tie. When asked about likelihood of continuing to use the BTAT, 35% were undecided and 53% planned to continue to use it.

The final two questions of the follow-up test were free text and focused on overall understanding and thoughts pertaining to tongue-tie; the results are listed in Table 6. Twenty-six percent (4 people) said they had no change in their understanding or management of tongue-tie. Respondents reported being able to more objectively assess for tongue-tie, being unsure about the efficacy of frenotomies, having more knowledge about tongue-tie and having not encountered a tongue-tie since the educational in-service. The most intriguing comment was that “your talk brought it [tongue-tie] from the realm of magic to something that is amenable to rational decision”.

Table 6

Final comments on understanding/management and thoughts

<p>Has your understanding or management of tongue-tie changed in the past six weeks?</p>	<p><u>Increased objective assessments</u></p> <ul style="list-style-type: none"> • Yes, better able to quantify using scale • Yes. I feel like I now have a less subjective way to assess the possible benefit of frenotomy for my patient • I may be more inclined to support treatment of tongue tie if indicated by the BTAT scores and based on presentation <p><u>Increased knowledge</u></p> <ul style="list-style-type: none"> • My understanding has but I have not had to put it into practice • I gained more knowledge from the in-service and definitely feel more equipped. I have not had the opportunity to use it yet, just because I do not do primary care much! • I am more aware of it now than before • Yes. Your talk brought it from the realm of magic to something that is amenable to rational decision
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	<p><u>Skepticism</u></p> <ul style="list-style-type: none"> • Less convinced regarding efficacy of procedure <p><u>No change</u></p> <ul style="list-style-type: none"> • No (reported by 3 participants) • No, unfortunately I have not encountered a tongue-tie <p><u>Other</u></p> <ul style="list-style-type: none"> • I like the evaluation tool, but I did not get any follow up from the patients (I had only 2 during this time period) • Yes • To the extent of breastfeeding issues, referral and lack of support
<p>What additional thoughts do you have about tongue-tie?</p>	<p><u>Skepticism</u></p> <ul style="list-style-type: none"> • I think the lactation consultants “accuse” tongue of causing problems more often than necessary and some parents then have excessive worry about that • I was taught that if the tongue could get over the gum line, it would cause no problems. Anecdotally I have seen kids with tight tongue ties that had no difficulty breastfeeding and seen kids who have had their tongue and lip ties cut with no improvement in breastfeeding. I am not a fan of frenulotomies. • Even after studies reviewed I still do not believe that upper lip tie is a significant cause of breastfeeding issues or speech issues. Lacking supportive evidence. • It is still probably over treated <p><u>Other</u></p> <ul style="list-style-type: none"> • A poor variant in our biological evolutionary process. Would be interesting to measure the incidence of tongue tie in third world countries • Considering tongue tie is valuable when evaluating patients with difficulty feeding

Discussion

Of all eligible participants, the greatest participation of providers was by pediatricians. Eight-five percent (n=11) of pediatricians employed at After Hours Pediatrics participated while only 22% (n=2) of physician assistants, 44% (n=4) of nurse practitioners and 60% (n=3) of nurses participated. Initially this seems surprising as previously reported surveys of providers found pediatricians to be more skeptical about ankyloglossia. It likely was related to convenience as the educational in-services were offered during the lunch break at midday. The pediatricians work standard 8am-5pm schedules while the majority of physician assistants and

nurse practitioners work alternate schedules which include some day hours but many evenings and weekends. Half of the nurse practitioners and physician assistants who did participate came to educational in-services that were held on their days off, while no pediatricians attended on days off.

The educational in-service appeared to have minimal effects on some aspects of provider perception of tongue-tie. For example, the responses changed minimally in the two post-tests for the question about how often does tongue-tie effect breastfeeding. None of the respondents were undecided by the follow-up test. An initial increase in negative effects of tongue-tie on breastfeeding was seen in the posttest, but by the follow-up test the responses were very similar to the pretest.

Other aspects of provider perception changed as a result of the educational in-service. Fewer providers felt that frenotomies were beneficial for breastfeeding. Initially, 60% believed they were beneficial but this dropped to 34%; the percentage who answered undecided increased from 25% to 50%. Provider perception of negative side effects also changed with zero providers stating there were negative side effects initially to one believing there are negative side effects and two who were undecided. The educational in-service focused on reviewing the five randomized controlled trials done to date on ankyloglossia along with several other more recent studies that were pertinent. As discussed within the review of literature, there are overall few studies that address ankyloglossia, and the ones that do exist tend to have small sample sizes with many methodological and reporting issues. Most of the participants had not previously reviewed the studies and were not impressed by them. The lack of high quality and robust research on the topic likely led to fewer providers believing that frenotomies are beneficial.

Several participants discussed physical qualities of the mouth when asked about assessing

for tongue-tie in the pretest. The most common response related to assessment parameters was tongue protrusion followed by assessing the frenulum, tongue shape, nipple pain and then breastfeeding. The BTAT tool assesses tongue tip appearance, attachment of the frenulum to the lower gum ridge, lift of the tongue with the mouth wide, and protrusion of tongue. The only aspect of the BTAT not mentioned at all by participants was lift of the tongue. No participants discussed objective measures. Only 24% (four respondents) discussed the use of the BTAT as an indicator for frenotomy in the posttest and no other objective measures were mentioned. No participants reported objective data or improvement within the BTAT scores as desired outcomes for frenotomies. The educational in-service did discuss numerous measurement tools for breastfeeding and latch quality/efficacy that were used in previous studies. Additionally, the purpose of this quality improvement project was to standardize assessment and management of ankyloglossia within a pediatric practice through the introduction and collective use of the BTAT. The low percentage who even mentioned the use of the BTAT as an indicator for frenotomy therefore was disappointing.

While 84% of respondents in the posttest thought of the BTAT as user friendly, there was less enthusiasm for its use. Some of this may stem from general lack of belief in the efficacy of frenotomies and of ankyloglossia's effect on breastfeeding. While 36% of respondents had encountered tongue-tie during the six week follow up period, only 18% reported having used the BTAT. At the six week follow-up test, only 77% agreed that the BTAT was easy to use. Approximately half of providers found the BTAT tool helpful at facilitating conversations with parents and guiding clinical decisions regarding management. Only 53% of providers planned to continue using the BTAT, although 35% were undecided and could possibly be swayed.

Despite the lack of strong research, some participants were willing to learn how to perform frenotomies. In the pretest, only 13% (2 people) reported performing them. This is under

reported as at least four of the participants during discussions at the educational in-services discussed their personal experiences with the procedure. The last four questions on the pretest, including the question about having had performed frenotomies previously, were located on the back page and required turning the page over. Four people missed all four questions; they likely did not notice the second page. Therefore, a more accurate assumption is that 20% of providers surveyed had previously performed frenotomies. In the pretest 57% of respondents were willing to perform frenotomies if trained.

Much skepticism about tongue-tie and frenotomies remained. Throughout the free text questions on all three tests, respondents wrote about lack of evidence to support frenotomies, frenotomies generally not being needed, feeling that the pediatric dental community was monetizing tongue-tie, thinking that reported improvements were due to parental perception or placebo effect, that lactation consultants over focus on tongue-tie and cause unneeded worry in parents, and that tongue-tie is over treated. Approximately half of the providers that encountered tongue-tie during the follow up period used the BTAT tool. In the follow-up test many comments were shared that expressed skepticism. It is unlikely that pediatric clinics will adopt widespread use of the BTAT tool without more rigorous studies supporting its use and the efficacy of frenotomies for tongue-tie.

This was the first survey of provider perception of tongue-tie that included physician assistants, nurses and nurse practitioners. A higher percentage of respondents felt that tongue-tie negatively impacted breastfeeding than compared to pediatricians within Messner and Lalakea's 2000 study. There were too few participants to be able to compare responses based on type of provider. Additionally, doing so would have compromised anonymity. In the Brinkmann et al. (2004) survey of Australian surgeons, most surgeons felt that frenotomies did help improve breastfeeding. While a slight majority of participants (60%) felt frenotomies improved

breastfeeding initially, only 34% believed so following the educational in-service. Similar to the Brinkmann et al. (2004) study, there were no universally agreed upon ways to assess for ankyloglossia, specific indications for frenotomies, or consistent expected outcomes. Even in the posttest, only four people discussed use of the BTAT tool as an indication for frenotomies. There were themes in expected outcomes such as improvements in breastfeeding, nipple pain, weight gain and tongue protrusion.

Implications for practice

One of the objectives of this quality improvement project was to standardize assessment and management of ankyloglossia within a pediatric clinic. This was partially successful with some providers incorporating the use of the BTAT into their clinical practice. Previously, none of the providers were even aware of the BTAT. The BTAT was chosen for the educational in-service as it was the most user-friendly tool, had proven reliability and validity, and helped to create an objective way to assess and manage ankyloglossia that took away some of the guess work and individual biases. Approximately half of the providers who attended the educational in-service planned to continue using the BTAT. Having half of the providers managing ankyloglossia in a standardized manner is a small victory, although there is room for greater uptake of the use of the BTAT as a diagnostic tool for ankyloglossia.

Gaining widespread buy in of using the BTAT may be challenging. The educational in-service did not greatly change provider perception of the effect of tongue-tie on breastfeeding. Much skepticism about tongue-tie remained. Most providers had not previously examined the literature on the topic. Numerous comments on open ended questions on the tests and conversations during the educational in-services focused on the lack of robust studies on tongue-tie. With lack of robust studies many providers did not want to change their current practices. Additionally, many comments included thoughts about tongue-tie being over diagnosed/treated, monetized by dentists, and frenotomies only having placebo effects showed that much

skepticism remains.

The goal of standardizing assessment and management of ankyloglossia is to provide reassurance to families whose infants are less likely to benefit from a frenotomy, and avoid an unnecessary procedure, while offering frenotomies to infants who are most likely to benefit from them. The vast majority of providers found the BTAT tool easy to use. More widespread incorporation of the tool into practice would improve the diagnosis of ankyloglossia, and ideally actually reduce the total number of frenotomies being done within the community. This was demonstrated in the Dixon et al. (2018) study. Additionally, half of providers found the use of the BTAT helped to facilitate conversations with families about ankyloglossia and helped guide clinical decisions. Only about one third of providers actually encountered tongue-tie in the follow up period. It is likely that with a longer follow up period, more would have encountered tongue-tie and more would have reported the tool useful for facilitating conversations and guiding clinical decisions.

Further incorporation of the BTAT by all professionals who interact with infants, and especially those who assist with breastfeeding, would help to minimize the conflicting advice that families often receive. Further research on the BTAT tool and ankyloglossia is needed. Possibly with more robust studies more professionals will be willing to incorporate the BTAT into their practice.

Limitations for health policy

There have been many healthcare policies that have focused on and promoted breastfeeding. Policies that promote breastfeeding are likely to continue with the current national and international focus on breastfeeding as the optimal nutrition for babies (U.S. DHHS, 2011). Therefore, anything that can facilitate breastfeeding should also be encouraged, such as helping with management of ankyloglossia. Policies that help increase funding for further research on

ankyloglossia could be one starting point. Potentially with more robust research, professional organizations may create practice guidelines for ankyloglossia. Without further research, implications of health policies pertaining to ankyloglossia are somewhat limited.

Limitations and strengths of the study

This was the first time that perceptions about tongue-tie of physician assistants, nurse practitioners and nurses were explored. The response rates for the posttest and follow-up test were high. Many of the free text questions allowed for greater insight into the perspectives of participants. Limitations included being an overall small sample with only twenty participants, representing only half of the eligible participants at the study site. Due to the small size of the sample, results are not generalizable, although many responses were similar to the two previous studies that were done on provider perception. The lack of high-quality studies on ankyloglossia detracted from the quality of the shared research, and likely contributed to skepticism on the topic. Even the research on the BTAT that was presented is deficient, without an identified exact score to be definitively used to recommend frenotomies.

One major goal of this project was to standardize the assessment and management of ankyloglossia through clinic wide incorporation of the BTAT in newborn visits. Because only about half of participants plan to continue its use, much of the actual management of ankyloglossia within the clinic will remain subjective and based on individual providers discernment allowing for more bias. Many providers did not feel frenotomies helped breastfeeding or that tongue-tie significantly impacted breastfeeding after the educational in-service. Interestingly, widespread incorporation of the BTAT would likely decrease overall rates of frenotomies as it did in the Dixon et al. (2018) study.

Suggestions for further research

Further research on ankyloglossia is clearly needed; the studies done to date have many methodological flaws and often very small sample sizes. Higher quality studies, and ideally, randomized controlled trials with larger sample sizes that seek to control for numerous biases and include true control groups are needed that explore the actual effectiveness of frenotomies. Further research using the BTAT is necessary to identify a definitive score at which point frenotomies are recommended. Without a universally agreed upon definition of ankyloglossia, true prevalence rates are unknown. During the educational in-services several participants had asked about prevalence rates in developing countries. If ankyloglossia truly is so prevalent and can affect breastfeeding significantly, one would expect to see similar issues in more areas without universal access to care. No such studies have been conducted. Further research into provider perception of ankyloglossia, specifically comparing in-hospital vs. clinic providers would also provide another perspective. There is even less research available on posterior tongue-tie and labial frenulums, yet some professionals who conduct frenotomies recommend also releasing these. Additionally, ideal timing for frenotomies has yet to be established. Much research is needed still. With more robust studies, health professionals would have increased confidence in making clinical decisions and recommendations for families of infants with ankyloglossia grounded in evidence-based research.

Conclusion

Diagnoses and treatment of ankyloglossia has been noted to be increasing (Walsh et al., 2017). And many more families are asking about it. With national and international efforts to promote breastfeeding as the optimal nutrition for infants, the focus on ankyloglossia is likely to increase with time. Providers need to be informed on the topic and know how best to help families. With limited high-quality studies on ankyloglossia, many providers may find the topic confusing.

Frenotomies have often been sought out by parents, possibly based on poor advice. Providers need to be prepared to address the topic knowledgeably and thoughtfully. One approach would be to incorporate the BTAT into clinical practice to help with encouraging frenotomies for infants most likely to benefit from the procedure while reducing unnecessary procedures for those less likely to benefit; this may lead to an overall decrease in unnecessary frenotomies. Further research on the topic is needed, although providers must be prepared to address it, ideally from an objective perspective based on current evidence-based research.

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Appendix A. Bristol Tongue Assessment Tool

Table 1 Bristol Tongue Assessment Tool (BTAT)				
	0	1	2	Score
Tongue tip appearance	Heart shaped	Slight cleft/notched	Rounded	
Attachment of frenulum to lower gum ridge	Attached at top of gum ridge	Attached to inner aspect of gum	Attached to floor of mouth	
Lift of tongue with mouth wide (crying)	Minimal tongue lift	Edges only to mid-mouth	Full tongue lift to mid-mouth	
Protrusion of tongue	Tip stays behind gum	Tip over gum	Tip can extend over lower lip	

(Ingram et al., 2015)

- In the past six weeks have you encountered newborns with tongue-tie? (yes/no)
- For infants with tongue-tie, how often have you used the Bristol Tongue Assessment Tool (BTAT)? (Likert Scale, NA)
- Have you found the BTAT easy to use? (Likert Scale, NA)
- Has the use of the BTAT helped facilitate conversations with parents about tongue-tie? (Likert Scale, NA)
- Has the use of the BTAT helped you make decisions about management of tongue-tie? (Likert Scale, NA)
- How likely are you to continue using the BTAT tool for assessment of tongue-tie? (Likert Scale)
- Has your understanding or management of tongue-tie changed in the past six weeks? (free text)
- What additional thoughts do you have about tongue-tie? (free text)

Appendix D. Educational In-service Outline

- Pre-test questionnaire
- Power point presentation
 - Introduction and background
 - Objective assessment tools for ankyloglossia
 - Focusing on the Bristol Tongue Assessment Tool (BTAT)
 - Randomized controlled trials
 - Recent research
 - Posterior ankyloglossia and labial frenulums
 - Ideal timing of frenotomies
 - Frenotomy revision
 - Laser vs scissors
 - Provider perception
 - Community efforts to decrease frenotomies
 - Strengths/limitations of current research
- Questions/clarification of the BTAT tool
- Post-test questionnaire

Research Data and Study Records

Researchers and staff should have systems or practices for maintaining the essential Research Records that they create in order to be able reasonably to support research findings, justify the uses of research funds and resources, and protect any resulting intellectual property.

During the life of a study and beyond its closure, many information security and storage policies pertain to the maintenance and archival of study documents and research data. These policies and procedures include those of the researcher's department, UNM HSC, the State of New Mexico, Federal privacy laws (such as HIPAA, FERPA, FOIA, New Mexico IPRA), Federal regulations (FDA, OHRP, DHHS, etc) as well as the data confidentiality requirements associated with research funding (e.g. National Institutes of Health, Department of Defense (DOD), etc.)

PI responsibilities for document and data security are particularly critical during times of study transition, as when a PI is leaving UNM HSC, is transferring PI responsibilities or is closing a study. Be prepared ahead of time and discuss transition and/or long-term storage plans with your department Chair/ Research Chair. Assure that information regarding these plans are documented in a standard place and are using an established process, so that an incoming PI and department personnel can find, understand and follow it.

Appendix A-1 Reportable New Information

Report information items that fall into one or more of the following categories to the HRRC within 5 business days. Reference SOP: New Information (HRP-024).

- 1) Information that indicates a new or increased risk, or a new safety issue, for example:
 - a. New information (e.g., an interim analysis, safety monitoring report, publication in the literature, sponsor report, or investigator finding) indicates an increase in the frequency or magnitude of a previously known risk, or uncovers a new risk.
 - b. Protocol violation that harmed participants or others or that indicates participants or others might be at increased risk of harm
 - c. Complaint of a participant that indicates participants or others might be at increased risk of harm or at risk of a new harm
 - d. An investigator brochure, package insert, or device labeling is revised to indicate an increase in the frequency or magnitude of a previously known risk, or describe a new risk
 - e. Withdrawal, restriction, or modification of a marketed approval of a drug, device, or biologic used in a research protocol
 - f. Changes significantly affecting the conduct of the clinical trial or increasing the risk to participants
- 2) Harm experienced by a participant or other individual, which in the opinion of the investigator are unexpected and *related or possibly related* to the research procedures.
 - a. A harm is "unexpected" when its specificity or severity are inconsistent with risk information previously reviewed and approved by the HRRC in terms of nature, severity, frequency, and characteristics of the study population.
 - b. A harm is "related or possibly related" to the research procedures if, in the opinion of the investigator, the research procedures more likely than not caused the harm.

- 3) Non-compliance with the federal regulations governing human research or with the requirements or determinations of the HRRC, or an allegation of such non-compliance
- 4) Failure to follow the protocol due to the action or inaction of the investigator or research staff
- 5) Change to the protocol taken without prior HRRC review to eliminate an apparent immediate hazard to a participant
- 6) Breach of confidentiality
- 7) Complaint of a participant that cannot be resolved by the research team
- 8) Premature suspension or termination by the sponsor, investigator, or institution
- 9) Incarceration of a participant in a study not approved by the HRRC to involve prisoners
- 10) Audit, inspection, or inquiry by a federal agency and any resulting reports (e.g., FDA Form 483)
- 11) Written reports of study monitors
- 12) Unanticipated adverse device effect (any serious adverse effect on health or safety or any life-threatening problem or death caused by, or associated with, a device, if that effect, problem, or death was not previously identified in nature, severity, or degree of incidence in the investigational plan or application (including a supplementary plan or application), or any other unanticipated serious problem associated with a device that relates to the rights, safety, or welfare of participants)
- 13) Unanticipated Problems Involving Risks to Subjects or Others, including any event or problem that is serious, unexpected, and related to the research, where “related” means the event or problem might reasonably be regarded as caused by, or probably caused by, the research.
- 14) Disciplinary action against the investigator or research staff by federal, state, and local regulatory agencies.