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A pilot study of non-nutritive suck measures immediately pre- and post-frenotomy in full term infants with problematic feeding

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ABSTRACT

Introduction: The purpose of this study was to describe symptoms of problematic feeding in infants with tonguetie, evaluate changes in non-nutritive suck measures before and after frenotomy, and examine tongue-tie severity with changes in non-nutritive suck patterning.

Method: Parents completed the Neonatal Eating Assessment Tool about infant feeding before frenotomy. Nonnutritive suck data were collected for 5 min before and after frenotomy. We used paired t-tests to compare non-nutritive suck measures pre- and post-frenotomy and linear regression evaluated the effect of tongue-tie severity and infant behavioral state on change in non-nutritive suck mechanics.

Results: Twenty-one infants had scores that met criteria for problematic feeding. The infant's non-nutritive suck amplitude (cm H_2O) (p=.02) and non-nutritive burst duration (sec) (p=.03) decreased post-frenotomy.

Discussion: This study supports the need for additional research to better understand feeding problems and changes in non-nutritive suck amplitude and duration in infants with tongue-tie.

1. Introduction

Tongue-tie is a mild congenital anomaly, described as a midline fold (Mills et al., 2019), that restricts tongue motion and function. Restricted tongue mobility may alter latch onto the breast or bottle nipple and interfere with effective swallowing (Geddes et al., 2008). Professional consensus recognizes that tongue-tie may be a cause of breastfeeding problems (Messner et al., 2020), namely nipple pain, and may be a reason for early cessation of breastfeeding (Ricke et al., 2005). Concurrent lip tie is commonly treated, but it has not been determined to be a source of breastfeeding problems (Messner et al., 2020). Prevalence of tongue-tie is documented as 8% in children less than one year of age (Hill et al., 2020). Treatment for tongue-tie is frenotomy, and this procedure has grown in popularity in the last 20 years, with an 866% increase in frenotomy rates in infants since 1997 (Walsh et al., 2017).

Milk flow from the breast to the oral cavity of the infant corresponds to downward movement of the infant's tongue and vacuum pressure during sucking (Geddes et al., 2008). This suggests that suck strength is an integral component of milk removal while breastfeeding. Ultrasound imaging has documented tongue movements necessary for milk removal, with impairment in both tongue motion and peristalsis in infants with

tongue-tie. Two distinct tongue movements have been observed in infants with tongue-tie that either compress the nipple base or increase the distance between the hard soft palate junction (HSPJ) and the maternal nipple (Geddes et al., 2008). In both types of observations, maternal nipple pain or damage can occur as the infant may bite rather than suck on the nipple or fail to sustain an adequate seal onto the breast during feeding. Milk-transfer rates and milk intake, measured by the test-weigh measure, improved following frenotomy in a small sample in prior research (n = 8 and 6, respectively) (Geddes et al., 2008). In other words, the infant's weight was significantly higher after a feeding, demonstrating a greater amount of milk extracted from the breast following tongue-tie correction. Tongue and lip motility have been studied using ultrasound in a case report of one infant with tongue-tie and one infant with lip-tie before and after frenotomy (Genna et al., 2021). The results demonstrated improvements in tongue motility. Specifically, the infant with tongue-tie demonstrated chaotic and unstable periodicity, with improvements that coincided with normal infant anatomy immediately post-frenotomy.

It remains unclear which infants benefit most from frenotomy due to the absence of high-level evidence and variability in screening and diagnostic criteria (O'Shea et al., 2017). Moreover, most available

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research focuses on maternal breastfeeding symptoms in the setting of tongue-tie (Hill and Pados, 2020). A recent study found that symptoms of problematic feeding are significantly reduced following correction of severe (Class III) tongue-tie (Hill et al., 2021). Physiologic data to assess how infant feeding mechanics improve following frenotomy have not been evaluated beyond studies with small samples sizes, lacking adequate power for statistical analyses, limiting generalizability of findings (Geddes et al., 2008; Genna et al., 2021). Most research on tongue-tie has involved subjective visual examination (Andrade Campanha et al., 2019) and assessment of maternal report of breastfeeding problems (Hill and Pados, 2020). There have been no studies to date that examine objective and comprehensive sucking measures in the setting of tongue-tie, and how these may change after correction via frenotomy. Non-nutritive sucking (NNS), defined as sucking without the provision of nutrients (Humphrey, 1970), is a precursor to oral feeding and has been used to improve our understanding of oral feeding in premature infants (Bingham et al., 2010; Pineda et al., 2019), development of physiologic stability (Foster et al., 2016), and future language development (Wolthuis-Stigter et al., 2017). Non-nutritive sucking is organized into bursts of sucking and pause periods for respiration. Each burst contains approximately 6-12 cycles (Wolff, 1968). Prior work has shown that NNS can influence behavioral state in infants (Dipietro et al., 1994), especially when it occurs before a feed (Gill et al., 1992; McCain, 1995). NNS has also been measured in a variety of infant populations with prior literature characterizing NNS across populations with a variety of clinical diagnoses (da Costa et al., 2010; Estep et al., 2008; Poore et al., 2008).

Thus, the purpose of this study was to improve our understanding of the impact of tongue-tie on infant feeding before correction and on NNS mechanics before and after frenotomy. Specifically, we aimed to describe problematic feeding symptoms before frenotomy; evaluate changes in NNS measures pre- to post-frenotomy; and examine the impact of tongue-tie severity and infant behavior on changes in NNS mechanics. We hypothesized that NNS would be altered post-frenotomy and that infants with more severe tongue-tie would have the greatest change in NNS measures following frenotomy.

2. Method

Institutional Review Board approval was obtained by the university. Participants were provided a comprehensive consent form and agreed to participate before data collection.

2.1. Design

This was a one-group cross-sectional pre/post intervention study design.

2.2. Setting

This study took place at a dental office in the northeast region of the United States between November 2020 and February 2021. There is one dentist at the location who conducts all exams using Kotlow diagnostic criteria (Kotlow, 1999) and an anatomical/functional scoring system. The dentist or parent held the infant during the NNS assessments following simple instructions from a trained researcher.

2.3. Sample size calculation

In prior studies utilizing NNS, there was ample power for statistical analysis in a sample of 12 infants. As this was a pilot study, an a priori power analysis was not completed. We chose a sample size of 25 mother-infant dyads.

2.4. Sample

To be included, infants had to be less than 7 months old, diagnosed with tongue-tie and undergoing frenotomy. A cutoff age of 7 months was selected to reduce variability with older infants when solid foods are introduced around this age and the maturational changes that occur over time (Wilson et al., 2008). Mothers were required to be 18 years of age or older and proficient in English to consent, read, and answer survey questions. Exclusion criteria were infant prematurity (less than 37 weeks' gestation at birth) (Pineda et al., 2019; Zimmernan and Rosner, 2018) and other anomalies that impact feeding (e. g., cleft lip or palate) (Reid, 2006). All eligible mother-infant dyads who presented to the office during the time of the study were invited to participate.

2.5. Frenotomy intervention

Assessment by the dentist and determination of need for frenotomy was conducted according to the dental office's standard practice and was not impacted by this research study. Informed consent for frenotomy was obtained by the dentist and treatment protocols were followed. Specifically, the infant's oral cavity was assessed for tongue, lip and buccal ties. If frenotomy was indicated, the infant was swaddled, eye protection was placed on the infant, and a dental hygienist immobilized the baby's head for the duration of the procedure. Anesthesia was not used. The tongue was lifted using a grooved-tip applicator and frenotomy was performed using a $\rm CO_2$ laser. For lip or buccal ties, the tissue was pulled away from the restriction and laser releases were performed (Figs. 1 and 2).

2.6. Data collection

Study data were collected and managed using REDCap (Research Electronic Data Capture) tools (Harris et al., 2009, 2019). Tongue-tie classification and anatomical/functional scores assigned by the dentist were obtained from the medical record. While the infant was undergoing frenotomy, the parent completed a survey using a tablet provided by the research team. The parent completed the Neonatal Eating Assessment Tool (NeoEAT) version (Breastfeeding, Bottle-Feeding, or Mixed Feeding) that coincided with the method(s) of feeding for the past seven days (Pados et al., 2018a, 2018b, 2019). Maternal and infant data were also obtained. Maternal data included pregnancy or birth complications, delivery mode, feeding plans prior to delivery, current feeding mode, maternal symptoms if breastfeeding, and feeding-related support within the last month (e.g., lactation consultant). We asked parents to select from the following maternal symptoms as applicable: creased, cracked or blanching nipples; painful latch onto the breast; gumming or chewing on the nipple; inability to latch; poor or incomplete breast drainage; infected nipples or breasts; abraded nipples; plugged ducts; mastitis; nipple thrush; over supply; under supply. Infant data included gestational age at birth, current medical diagnoses, medications used in the last week, and primary reason for dental evaluation. Infant feeding symptoms were ascertained from the parent via the NeoEAT. Type of bottle, bottle nipple, pacifier type and frequency of use was obtained, if applicable.



Fig. 1. Tongue-Tie* (*Photos in Fig. 1 and 2 owned and provided by pediatric dentist, used with permission).

R.R. Hill et al.



Fig. 2. Post-Frenotomy Release*. (*Photos in Fig. 1 and 2 owned and provided by pediatric dentist, used with permission).

2.7. Measures

2.7.1. Tongue tie diagnosis

Kotlow's criteria (Kotlow, 1999) was used to determine tongue attachment location, along with an anatomical/functional score of the infant's tongue. The Kotlow assessment involves measuring the length of the freely mobile tongue in millimeters. Classification ranges from least (Class I) to most restrictive (Class V). The anatomical/functional scoring system was adapted from the Hazelbaker Assessment Tool for Lingual Frenulum Function (ATLFF) scale (Hazelbaker, 1993). Scores range from 0 to 14 with higher scores indicating worse anatomical and/or functional impairment. Determination of need for frenotomy was made based on both of these measures as well as subjective concerns reported by the mother. Subjective concerns included pain with nursing, introduction of a bottle or supplementation with formula or expressed human milk due to breastfeeding difficulties. Prior to the visit, the dentist required an evaluation from a lactation consultant that indicated functional impairment with feeding, although this was not a component of this study's inclusion criteria.

2.7.2. Infant feeding

Symptoms of problematic feeding were assessed with the NeoEAT, a parent-reported measure of infant feeding that has been validated for use in infants less than seven months of age (Pados et al., 2017; Pados et al., 2018a; Pados et al., 2018b; Pados et al., 2019a; Pados et al., 2019b; Pados et al., 2020). Higher scores indicate more problems with feeding. The three NeoEAT versions (Breastfeeding, Bottle-Feeding, Mixed Feeding) include subscales that assess various aspects of feeding, such as energy and physiologic stability and gastrointestinal symptoms. All three versions of the NeoEAT have evidence of validity and reliability (Pados et al., 2019). Norm reference values have been developed (Pados et al., 2019a, 2019b, 2020). Problematic feeding is defined as a total score or any subscale score above the 90th percentile compared to the reference data.

2.7.3. NNS dynamics

The Non-Nutritive Suck (NNS) system evaluated infant suck measures. A custom portable NNS assessment cart was used to obtain suck samples from enrolled infants at each study session and has been used in prior studies (Heller et al., 2021; Martens et al., 2020; Zimmerman et al., 2020). The NNS system includes a pressure transducer attached to a Soothie pacifier (Philips Avent), a data acquisition system (Power Lab, ADInstruments), a laptop, and software (LabChart, ADInstruments). The device was calibrated before each day of data collection by calculating the percent error between pressure readings output from the NNS device's pressure transducer with a range of pressures input into the NNS device from a pressure calibrator. Infants sucked on the NNS device for 5 min each (pre- and post-frenotomy) and the two most active, consecutive minutes of the NNS sample, defined as the 2 min with the most NNS cycles, were used to calculate minute rate averages which were used for the present analysis. This is consistent with previous studies examining NNS (Barlow et al., 2008; Poore et al., 2008). For analysis, trained researchers utilized LabChart software to manually select and analyze NNS bursts using a custom Burst Macro. Bursts must contain two or more suck cycles, while the amplitude of bursts must be over $1 \, \mathrm{cm} H_2 0$, and a new burst begins when there is a break of more than 1000 ms between two cycles (Fig. 3). The NNS dynamics in this study included the following outcome measures: NNS cycles/burst, NNS cycles/minute, NNS amplitude (measured in cm $H_2 0$), NNS bursts/minute, NNS frequency (measured in Hz), and NNS burst duration (measured in seconds). The infant's behavioral state (e.g., asleep, quiet-awake, active crying) was documented for each NNS assessment.

2.8. Data analysis

All data were analyzed using SPSS v.25.. Sample characteristics were explained using descriptive statistics. For the NeoEAT, subscale and total scores were calculated for the appropriate version based on how the infant was fed in the prior week. Subscale and total scores were compared to reference data (Pados et al., 2019a, 2019b, 2020). Scores greater than the 90th percentile compared to reference data were defined as problematic. Internal consistency reliability of each of the NeoEAT scales for this sample was tested using Cronbach's alpha. Given the small number of infants with Class I and Class IV tongue-tie, the Kotlow score was recoded into a dichotomous variable; Class I or II and Class III or IV were grouped together for analysis. Infant behavioral state was also recoded into a dichotomous variable to indicate whether the infant was quiet or crying during NNS data collection before and after frenotomy.

For analysis of the NNS mechanics data, an initial analysis was run to identify outliers. Outliers were considered for removal if they significantly changed the results of the statistical test. If they did not significantly affect the results, they were kept in the analysis. Tests of normality were also examined to determine appropriate use of parametric tests. Paired t-test was used to compare NNS mechanics before and after frenotomy. Then, linear regression was used to evaluate whether there was a significant change in NNS mechanics, taking into account potential confounding variables. The effect of infant age in weeks, infant sex, and pacifier use on NNS mechanics were evaluated individually to determine whether these variables needed to be included in the model. Linear regression was then used to evaluate the effect of tongue-tie severity and infant behavior pre-frenotomy and postfrenotomy on the change in NNS mechanics (post-frenotomy score pre-frenotomy score). If found to be statistically significant in the individual model, infant age in weeks, infant sex, and/or pacifier use were also included. Statistical significance was defined as p < .05 for all tests.

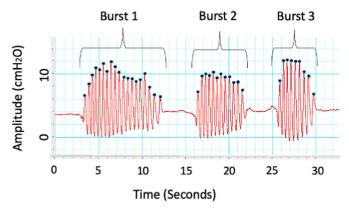


Fig. 3. An image of approximately 30 s of NNS activity for an infant enrolled in this study. Three NNS bursts are shown with black dots indicating suck cycle peaks.

3. Results

3.1. Sample characteristics

Twenty-five dyads participated in the study. Survey respondents were primarily the mother ($n=24,\,96\%$) of the infant and all were married, well-educated, with high socioeconomic status. The age of the parents ranged from 27 to 40 years ($M=32.6,\,SD=3.4$). Most mothers ($n=21,\,84\%$) planned to breastfeed the infant exclusively at the breast; two planned to breastfeed with pumped breast milk. Feeding plans changed for 11 (44%) of the dyads as a possible result of the infant's tongue-tie, with changes including adding pumped milk, bottle feeding, and initiating formula. Mothers reported several breastfeeding-related symptoms, the most common symptoms were difficult latching (71%), incomplete breast drainage (67%), and painful latching (63%). Twenty-three of the parents (92%) bottle-fed the infant previously, 12 of which (52%) noticed challenges with bottle-feeding. Parent demographics are presented on Table 1.

Infant age ranged from 1 week to 28.6 weeks (M = 9.1, SD = 7.7). All infants had tongue-tie, most diagnosed with a Class II (n = 11, 44%) or Class III (n = 12, 48%) severity score, indicating moderate or severe tongue restriction, respectively. The recorded anatomic-functional scores ranged from 9 to 13, indicating moderate to severe dysfunction. The caregivers saw an average of 2.2 providers, such as pediatricians and lactation consultants, prior to the infants' diagnosis of tongue-tie. In the month prior to frenotomy, nine of the infants (36%) received therapy to help with feeding and 16 infants (64%) received health care related to feeding issues such as visits for growth or feeding concerns, or reflux. Three infants had previously received a medical diagnosis of GERD, one requiring treatment with acid-reducing medication. Infants were otherwise healthy, with no other comorbidities noted. Eighteen infants (75%) routinely used a pacifier. Infant demographics are presented on Table 2.

Table 1 Demographics of participants (N = 25).

Characteristic	n (%)
Relationship to the Infant	
Mother	24 (96)
Father	1 (4)
Family Structure	
Two-Parent Family	24 (96)
Single-Parent Family	1 (4)
State of Residence	
Massachusetts	23 (92
New Hampshire	1 (4)
Rhode Island	1 (4)
Race	
White	24 (96
Asian	1 (4)
Ethnicity	
Not Hispanic or Latino	24 (96
Other (Polish American)	1 (4)
Highest Level of Education Obtained	
Associate Degree or Technical/Vocational/Occupational Program	1 (4)
Bachelor's Degree	12 (48
Master's Degree	11 (44
Doctoral Degree	1 (4)
Family Income in USD	
75,000–99,000	2 (8)
More than 100,000	23 (92
Providers that Participants Consulted Regarding Feeding-Related Issues	
Lactation Consultant	16 (64
Chiropractor	6 (24)
Pediatrician/Pediatric Nurse Practitioner	4 (16)
Craniosacral Therapist (CST)	3 (12)
Speech Therapist	1 (4)
Physical Therapist	1 (4)
Ears, Nose, and Throat Specialist (ENT)	1 (4)

Table 2 Demographics of infants (N = 25).

Characteristic	n (%)
Infant Sex	
Male	14 (56)
Female	11 (44)
Infant Age	
Less than 2 months	13 (52)
2-<4 months	9 (36)
4 months - less than 7 months	3 (12)
Race	
White	23 (92)
More than one Race (Asian and White)	2 (8)
Ethnicity	
Hispanic or Latino	1 (4)
Not Hispanic or Latino	23 (92)
Not Answered	1 (4)
Diagnoses	
Tongue-Tie	25 (100)
Lip Tie	19 (76)
Buccal Tie(s)	7 (28)
Ties Corrected	
Tongue-Tie	25 (100)
Lip Tie	18 (72)
Buccal Tie(s)	6 (24)
Kotlow Tongue-Tie Score	
1	1 (4)
2	11 (44)
3	12 (48)
4	1 (4)
Behavioral State Pre-Frenotomy	
Crying	2 (8)
Not Crying	23 (92)
Behavioral State Post-Frenotomy	
Crying	14 (56)
Not Crying	11 (44)

3.2. Infant feeding

Average NeoEAT total and subscale scores are presented on Table 3. Overall, 21 (84%) of the infants in this sample met criteria for having problematic feeding prior to frenotomy, as defined as either a total score or any subscale score on the NeoEAT being above the 90th percentile compared to a healthy, reference sample. All infants who were exclusively bottle fed (n = 3) or breastfed (n = 7) met criteria for problematic feeding and 11 of 15 (73%) infants who were fed by both breast and bottle met criteria for problematic feeding.

3.3. NNS dynamics

One infant's NNS cycles/burst and NNS burst duration were identified statistically as outliers and were removed from the analyses of these variables. After removal of these outliers, all NNS data were normally distributed (Shapiro-Wilks test p>.05). Results of the paired t-tests are presented on Table 4. NNS amplitude (t=2.45, p=.02) and NNS burst duration (t=2.25, p=.03) decreased significantly post-frenotomy.

Infant age, sex, or routine pacifier use were not significant predictors of change in NNS mechanics and were excluded from the linear regression models. There was no significant effect of tongue-tie severity or infant behavior pre-frenotomy or post-frenotomy on change in NNS cycles/burst, cycles/minute, bursts/minute, frequency, or burst duration. Infant behavior pre-frenotomy was a significant predictor of change in NNS amplitude (t=-5.48, p<.001), such that infants who were crying during NNS data collection pre-frenotomy had a significantly greater negative change in NNS amplitude.

4. Discussion

This study examined symptoms of problematic feeding before frenotomy and NNS patterning pre- and post- frenotomy in a cohort of

Table 3NeoEAT scores by subsample and presence of feeding concerns.

		Mean Score	SD	Score >90th PCTL n (%)
Breastfeeding (n = 7)				
,,	Total Score	116.1	17.5	3 (42.9)
	Infant Regulation	22.1	4.4	0 (0)
	Energy & Physiologic Stability	24.7	4.4	6 (85.7)
	Oro-Pharyngo-Esophageal Function	24.9	6.4	6 (85.7)
	Gastroesophageal Function	12.3	5	4 (57.1)
	Gastrointestinal Function	12.6	3.1	3 (42.9)
	Feeding Efficiency & Sensory Responsiveness	14.6	4.8	4 (57.1)
	Compelling Symptoms of Problematic Feeding	5	3.1	4 (57.1)
Bottle-Feeding (n = 3)				
	Total Score	113	13.9	1 (33.3)
	Infant Regulation	18.3	5.9	0 (0)
	Energy & Physiologic Stability	27	4.4	3 (100)
	Gastrointestinal Function	59.7	6	3 (100)
	Sensory Responsiveness	7	5.3	0 (0)
	Compelling Symptoms of Problematic Feeding	1	1	2 (66.7)
Mixed Feeding (n = 15)				
	Total Score	116.8	30	5 (33.3)
	Infant Regulation	18.7	5.8	0 (0)
	Energy & Physiologic Stability	24	11.1	10 (66.7)
	Gastrointestinal Function	47.9	15.9	7 (46.7)
	Sensory Responsiveness	7.1	5.5	2 (13.3)
	Feeding Flexibility	19.1	3.7	6 (40)

Note. PCTL = Percentile.

 $\label{eq:Table 4} \begin{tabular}{ll} \begi$

NNS Variable	Pre	Post	95% CI		t	p
	M (SD)	M (SD)	Lower	Upper		
Cycles/ Burst ^a	11.1 (6.21)	8.79 (4.28)	23	4.85	.47	.64
Cycles/Min Amplitude	46.42 (26.7) 13.52 (5.39)	44.02 (20.8) 10.25 (4.93)	−11.75 .51	16.55 6.04	.35 2.45	.73 .02*
Bursts/Min Frequency Duration ^a	4.66 (2.38) 2.08 (0.29) 5.21 (2.62)	5.1 (2.81) 2.11 (0.31) 4.04 (1.72)	-2.09 16 .09	1.21 .09 2.25	55 61 2.25	.59 .55 .03*

^{*}Indicates significance at the 0.05 level.

young infants. To our knowledge this is the first time both parent report of feeding and NNS physiology have been used together to examine sucking and feeding outcomes surrounding a frenotomy procedure.

In this sample, many parents reported significant symptoms of problematic feeding for their infants. Few studies have described symptoms of problematic infant feeding in infants with tongue-tie using a valid and reliable measure of feeding (Hill et al., 2021a,b). The findings from this study suggest that parents reported concerns of symptoms of problematic feeding in several areas, especially Energy & Physiologic Stability. This subscale of the NeoEAT examines the work of eating for infants, with questions such as "My baby is exhausted after eating" and "My baby can only suck a few times before needing to take a break." Future studies are needed to explore this in a larger sample and using additional measures to evaluate how effective the infant's suck is in

obtaining milk from the breast or bottle, if energy requirements for eating are reduced following tongue-tie correction, and if this corresponds with lower NNS amplitude.

There were concerning scores on the NeoEAT subscales that assess functions of the gastrointestinal tract for these infants. Prior research has demonstrated increased incidence of aerophagia-induced reflux in infants with tongue-tie (Siegel, 2017) and improvement in Gastroesophageal Reflux Disease (GERD) following frenotomy (Hand et al., 2020; Slagter et al., 2021; Ghaheri et al., 2017, 2018). In a sample of infants with tongue-tie, gastrointestinal tract symptoms assessed using the NeoEAT scales were significantly improved at the two-week follow-up after frenotomy (Hill et al., 2021a,b). Gastrointestinal and gastroesophageal symptoms were significantly reduced after frenotomy for younger infants and those with more severe tongue-tie when measured using a comprehensive GI symptom measure (Hill and Pados, 2021). Longitundal data is necessary to evaluate the effects of frenotomy on infant feeding and GI function over time.

There were significant changes in aspects of NNS physiology postfrenotomy procedure in NNS amplitude and NNS burst duration. These findings indicate that immediately after the procedure, NNS amplitude was signficantly lower and that NNS burst durations were significantly shorter. The exact reason for this NNS reduction in amplitude and burst duration remains unknown, and infant pain may have altered NNS measures immediately following frenotomy. The changes or lack of changes in NNS measures may have been impacted by infant pain immediately post-procedure, and infants with more oral restrictions (tongue, lip, and buccal) may have more significant pain. Prior work has shown that maturation, experience, and anatomical changes can alter NNS (Martens et al., 2020). Martens et al. (2020) showed that between 3 and 12 months of age all aspects of infants' NNS patterns changed significantly except NNS frequency. More specifically, compared to 3 months, 12-month-old infants has significantly higher amplitudes with shorter NNS burst durations. Taken together in the context of this study, perhaps the frenotomy procedure altered the anatomy to an extent that it changed the infants' suck amplitude and burst duration. It is positied that increased energy requirements with feeding may present as a higher sucking amplitude prior to frenotomy, as seen in this sample of infants. In light of the findings that infant behavior pre-frenotomy was a significant predictor of change in NNS amplitude, infant behaviors may be influencing this outcome.

Much of the prior work surrounding infant behavior and NNS has reported on the positive influence that NNS has on improved behavioral state (Dipietro et al., 1994; Gill et al., 1992; McCain, 1995). However, it is clear that crying during NNS will alter the NNS patterning. In our study, two infants were reported as crying during the pre-frenotomy data collection. One infant was soothed during the NNS procedure, and another was likely crying out of hunger as infants must remain NPO before the frenotomy procedure. Interestingly, tongue-tie severity did not significantly affect the change in NNS mechanics. However, the available assessment measures to diagnose and categorize tongue-tie do not have psychometric data available (Hill and Pados, 2020). Without this information, we do not know if the assessment modality used properly grades tongue-tie severity. More data are needed to examine these preliminary findings in more detail, ideally, the day before and the day and weeks after the procedure to reduce the influence of stress surrounding the procedure itself.

These data are among the first to examine parental report of problematic feeding as well as NNS metrics pre- and post- frenotomy procedure and have some clinical implications. The majority of parents reported problematic feeding prior to frenotomy, especially as it pertains to Energy & Physiologic Stability and Gastrointestinal Function. In addition, NNS burst duration and amplitude were lower post-frenotomy procedure. Taken together, these findings indicate that parents reported problematic feeding of their infant and that the frenotomy procedure can alter aspects of NNS. It is not clear at this time if parents feel that post-frenotomy their child's feeding is improved as we did not collect these

^a Analyzed without outlier (i.e., n = 24).

Journal of Neonatal Nursing xxx (xxxx) xxx

R.R. Hill et al.

data. It is also unknown how the altered NNS pattern post-frenotomy interacts with oral feeding success or growth metrics. Health care providers and developmental specialists should continue to use both parent report and physiological-based metrics to examine these important outcomes before and after tongue tie procedures, especially at different time points (e.g., the next day or week after procedure).

4.1. Limitations

The sample in this study was small and homogenous, representing mainly white infants with moderate to severe tongue-tie and many with concomitant lip-tie. The parents who participated were largely welleducated with high socioeconomic status. We cannot generalize these findings to the larger population, and do not know if the changes in NNS dynamics were due to the infant's tongue-tie, lip-tie, or a combination of the two. Future research would benefit from larger samples at multiple sites. Data collection over a longer span of time will help us to understand how problematic feeding symptoms and NNS dynamics change over time. Comparison of these variables in infants with tongue-tie with and without frenotomy would advance our knowledge of the utility of frenotomy and provide guidance on which infants benefit most from treatment. Lastly, though the dentist recommended parents not provide ibuprofen or other analgesia to infants until 4 hours after the procedure, we did not inquire whether infants were medicated by parents prior to procedure. This may have impacted the outcomes of the present study. Despite these limitations, this study is the first known to comprehensively evaluate both parent report of infant feeding and NNS metrics in infants with tongue-tie undergoing frenotomy, with clear directions for future research.

5. Conclusions

The majority of infants presenting for evaluation and correction of tongue-tie demonstrated concerning scores on the NeoEAT, a comprehensive measure of infant feeding. Following tongue-tie correction, there were significant decreases in NNS amplitude and NNS burst duration, regardless of infant age or tongue-tie severity. Larger studies with diverse samples and longitudinal evaluation of feeding and NNS metrics are needed to improve our understanding of this oral anomaly and its association with feeding and sucking dynamics.

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Role of funder/sponsor

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Author contributions

Rebecca R. Hill was responsible for the conceptualization, data curation, formal analysis, funding acquisition, investigation, methodology, project administration, supervision, and writing and revision of the manuscript.

Morgan Hines was responsible for the data curation, formal analysis, investigation, software, and writing and revision of the manuscript.

Alaina Martens was responsible for the data curation, formal analysis, investigation, software, and writing and revision of the manuscript.

Britt F. Pados was responsible for the conceptualization, formal analysis, methodology, resources, and writing and revision of the manuscript.

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Conflict of interest disclosures

The authors have no conflicts of interest to disclose.

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