


# Gastrointestinal Symptom Improvement for Infants Following Tongue-Tie Correction

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## Abstract

Treatment of tongue-tie in infants suggests improvements for breastfeeding mothers, including reduced nipple pain and improved latching onto the breast. The effects of frenotomy on infant feeding and gastrointestinal dysfunction remain controversial, with insufficient evidence on the relationship between tongue-tie and disorders of the gastrointestinal tract. The purpose of this study was to compare symptoms of gastrointestinal (GI) distress and gastroesophageal reflux (GER) prior to and 2 weeks following frenotomy in infants with tongue-tie. Parents were surveyed prior to frenotomy and 2 weeks post procedure, using the Gastrointestinal and Gastroesophageal Reflux (GIGER) Scale for Infants and Toddlers. Eighty-four participants completed surveys at both time points, with significant improvements in GI and GER symptoms 2 weeks after frenotomy. Younger infants and those with more severe tongue-tie had the greatest improvements in GI and GER symptoms. Infants with tongue-tie and symptoms of GI tract distress may experience improvement in symptoms after frenotomy.

## Keywords

ankyloglossia, gastroesophageal reflux, feeding behavior, infant

## Introduction

Ankyloglossia, more commonly known as tongue-tie, is an oral anomaly present in approximately 8% of infants.<sup>1</sup> This anomaly decreases tongue range of motion that can impair swallowing and eating. It is frequently associated with pain for the breastfeeding mother due to improper latch and increased friction of the tongue on the breast nipple.<sup>2</sup> Gastrointestinal (GI) distress can be the result of improper swallowing or excess ingestion of air (aerophagia). Although nonspecific, GI discomfort in infants can be distressing to parents. With the knowledge that acid-reducing medications have long-term negative sequelae for infants, including an increased risk of bone fractures,<sup>3</sup> it is important to identify possible causative factors of GI discomfort, such as tongue-tie. Few studies have evaluated gastroesophageal reflux disease (GERD) in infants with tongue-tie. Of those, findings suggest significant improvements in GERD symptoms following tongue-tie correction, almost immediately,<sup>4,7</sup> with continued improvements noted up to 6 months post procedure.<sup>8</sup>

Tongue-tie has been mentioned in literature dating back to the 1700s,<sup>9</sup> and was routinely treated up through the 1940s.<sup>10</sup> Around that time, infant formula became

more commonplace, the promotion of breastfeeding (BF) by health care providers declined, and screening for tongue-tie decreased.<sup>10</sup> Over the past half century, benefits of BF for both mother and baby have been identified,<sup>11</sup> with strategies from health care organizations aimed at increasing BF rates nationally and internationally. With this interest in increasing BF rates, there has been renewed interest in the contribution of tongue-tie to feeding challenges.

Research has noted improvement in GERD symptoms following tongue-tie correction through frenotomy.<sup>4,6,8</sup> These researchers all utilized the Infant Gastroesophageal Reflux Questionnaire–Revised (I-GERQ-R).<sup>12</sup> However, the I-GERQ-R has drawbacks for clinical use as it does not comprehensively evaluate GI and GER symptoms, and content validity has not been reported,<sup>13</sup> necessitating the use of a

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more comprehensive measure to examine GI tract symptoms in this population. During swallowing, the vocal folds and epiglottis close to protect the airway, and the base of the tongue contacts the posterior pharyngeal wall to create pressure to move the bolus downward (as opposed to moving up into the nasopharynx).<sup>14</sup> If the movement of the tongue is restricted, it may prevent the tongue from making appropriate contact with the posterior pharyngeal wall, resulting in nasopharyngeal reflux, and may interfere with the closure of the structures involved in protecting the airway, placing the infant at risk for aspiration. With impaired tongue movement, the infant may cough, make loud gulping sounds, or develop aerophagia.<sup>15</sup>

Treatment of tongue-tie is completed through frenotomy, using laser or scissors to release the excess frenum tissue beneath the tongue. Current practice for the treatment of tongue-tie is not standardized and insurance coverage for treatment is variable.<sup>16</sup> Despite this, a recent study reported an 866% increase in frenotomy rates from 1997 to 2012.<sup>17</sup> There are several classification systems to diagnose tongue-tie although none have been comprehensively assessed for validity and reliability. One of the more common assessment tools is Kotlow criteria. This method categorizes the tongue-tie from least (class I) to most (class IV) restrictive based on the amount of freely mobile tongue, measured in millimeters.<sup>18</sup> It is unknown which infants benefit most from frenotomy even with use of available diagnostic measures.

Using the theory of unpleasant symptoms<sup>19</sup> as the guiding theoretical framework, tongue-tie, as it relates to symptoms of GI and GER dysfunction, was explored. The theory of unpleasant symptoms is a middle-range theory designed to integrate the existing knowledge of symptoms that may guide future research and practice. The major tenets of this theory include the experience of symptoms, influencing factors that promote or alleviate symptoms, and the consequences associated with the symptoms.<sup>19</sup> Here, we discuss how tongue-tie contributes to GI and GER symptoms, and how these symptoms change following tongue-tie correction.

### **Purpose**

The purpose of this study was to explore changes in GI and GER symptoms for infants undergoing frenotomy before and 2 weeks following the procedure in a sample of infants less than 7 months of age. Our specific aims were to (1) explore changes in symptoms pre-frenotomy and post-frenotomy, (2) compare symptoms by tongue-tie severity and infant age, and (3) compare utilization of reflux strategies, both pharmacologic and nonpharmacologic, before and after frenotomy. It was hypothesized that GI and GER symptoms would decrease following

tongue-tie correction, and that infants with more severe tongue-tie would have greater improvements in symptoms after frenotomy. This study utilized a 1-group pre-intervention/post-intervention design, comparing symptoms before and after tongue-tie correction.

### **Sample**

Participants were recruited, using convenience sampling, from a pediatric dental office in the northeast. The pediatric dentist conducted the assessment for tongue-tie using Kotlow diagnostic criteria<sup>18</sup> and determined whether frenotomy was warranted, following their standard of practice. The standard care for the infant was not altered for this study. Once the infant was determined to need a frenotomy and the parent consented to the frenotomy procedure (using an isotopic CO<sub>2</sub> laser), the infant became eligible for this study. In addition, the infant had to be younger than 7 months of age, born full term (>37 weeks' gestation), and without other congenital anomalies (eg, cleft lip/palate) to be eligible. Parents needed to be 18 years of age or older and proficient in English to provide consent and to participate. All individuals who met these inclusion criteria were invited to participate between July and November 2020. The total sample for this study was 84 parent-infant dyads.

### **Methods**

This study was approved by the institutional review board at the university. A convenience sample was recruited between July and November 2020. Eligible parent/infant dyads were identified by a member of the pediatric dental staff and were provided with information about the study. Informed consent was obtained through an online consent form prior to the start of the online survey. The principal investigator was available to answer questions about the study prior to commencement. Data for the first survey were collected on a password-protected and encrypted electronic tablet provided by the research team on the day of the dental visit. The follow-up survey was sent to participants 2 weeks post-frenotomy through email. Medical record data were provided by the dental office staff to the principal investigator, stored on a password-protected and encrypted laptop.

Study data were collected and managed using REDCap (Research Electronic Data Capture) electronic data capture tools.<sup>20,21</sup> REDCap is a secure, web-based software platform designed to support data capture for research studies, providing (1) an intuitive interface for validated data capture, (2) audit trails for tracking data manipulation and export procedures, (3) automatic export procedures for seamless data downloads to

**Table 1.** Description of GIGER Subscales.

GIGER subscale	Purpose	Exemplar
Difficulties with self-regulation	Reflects infant's ability to regulate during eating	"My child is calm and relaxed when eating." "My child needs to be encouraged to keep eating (such as by touching or talking)."
Common GI and GER symptoms	Concerns suggesting less severe, more common GI and GER symptoms	"My child is uncomfortable if laid flat after eating." "My child spits up in between feedings."
Compelling GI and GER symptoms	Concerns suggesting more severe GI and GER symptoms	"My child stops breathing or struggles to breathe during feedings/mealtime." "My child has blood or mucous in poop/stool."

Abbreviations: GER, gastroesophageal reflux; GI, gastrointestinal; GIGER, gastrointestinal and gastroesophageal reflux.

common statistical packages, and (4) procedures for data integration and interoperability with external sources.

At both time points, participants completed the 36-item Gastrointestinal and Gastroesophageal Reflux (GIGER) Scale for Infants and Toddlers. The GIGER scale is a parent-reported measure of symptoms of GI distress and GER. The GIGER has evidence of adequate psychometric properties, including acceptable internal consistency reliability (Cronbach  $\alpha = .78-.94$ ), convergent validity, and known-groups validity (Pados et al., 2021).<sup>13</sup> This measure rates symptoms on a 6-point Likert scale ranging from *never* to *always* in 3 domains, Difficulties with Self-Regulation (reverse-scored), Common GI and GER Symptoms, and Compelling GI and GER Symptoms. Higher scores indicate more symptoms of GI and GER dysfunction; scores can range from 0 to 180. Examples of questions on the GIGER are presented in Table 1. In addition to the GIGER, parents were asked whether their infant had received a formal diagnosis of GERD from a health care provider, whether they believed their infant was experiencing symptoms of GER, and whether they were using nonpharmacologic (eg, paced feeding, holding upright after feeding, or elevating head of bed) or pharmacologic (eg, proton-pump inhibitors) strategies to manage infant reflux.

### Data Analysis

An a priori power analysis was conducted using G\*Power 3.1.9.4 (G\*Power, Germany). The power analysis determined that a sample size of 53 was required to obtain 95% power with a medium effect size (0.5) for 2-tailed significance at .05.

The sample was described using descriptive statistics. Reliability analyses were completed for the GIGER scale using Cronbach alpha. Tests of normality were run for the total and subscale scores of the GIGER pre-frenotomy and post-frenotomy, revealing non-normal

distribution of the data. Nonparametric tests (Wilcoxon signed-rank test) were run to examine differences in mean GIGER scores pre-frenotomy and post-frenotomy. To evaluate the effect of infant age and tongue-tie severity on the changes in GIGER symptoms, a change score was calculated for the total and subscale scores by subtracting pre-frenotomy from post-frenotomy scores. We used linear regression to determine the effect of infant age (in weeks) and tongue-tie severity (Kotlow category I-IV) on change score for GIGER total score and for each of the 3 subscale scores. Logistic regression was used to explore changes in reflux strategies and parental perception of reflux symptoms pre-frenotomy and post-frenotomy. Poisson regression was used to determine whether there were significant changes in the number of strategies to manage reflux pre-frenotomy to post-frenotomy. Statistical significance was set at  $\alpha = .05$  (2-tailed) for all statistical tests.

### Results

Parental age ranged from 19 to 41 years (mean = 31.5, SD = 4.9) and infant age ranged from 0.4 weeks to 29.4 weeks (mean = 6.3, SD = 6.4). Respondents were mainly Caucasian and non-Hispanic and married. All infants were diagnosed and treated for both tongue and lip ties, with no complications noted immediately following the procedure. Most infants ( $n = 75$ ) were diagnosed with a Class III tongue-tie. Full demographic information is presented in Tables 2 and 3.

Reliability of the GIGER scale was excellent pre-frenotomy and post-frenotomy ( $\alpha = .92$  and  $.87$ , respectively). Mean GIGER total score was 51.88 pre-frenotomy and 37.94 post-frenotomy, representing a statistically significant improvement in GER and GI symptoms following tongue-tie correction ( $P < .001$ ). All 3 subscales had significantly lower scores post-frenotomy ( $P < .001$ ). Table 4 depicts total and subscale scores for the GIGER at both time points.

**Table 2.** Demographics of Participants (N = 84).

Characteristic	No. (%)
Relationship to infant	
Mother	78 (92.8)
Father	5 (6)
Other (cousin)	1 (1.2)
Race	
American Indian/Alaskan Native	1 (1.2)
Asian	1 (1.2)
Black/African American	1 (1.2)
White	79 (94)
More than 1 race	1 (1.2)
Other	1 (1.2)
Ethnicity	
Hispanic/Latino	2 (2.4)
Not Hispanic/Latino	75 (89.3)
Other	4 (4.8)
Unknown	3 (3.5)
Education level	
High school/GED	15 (18)
Associate's degree	16 (19)
Bachelor's degree	27 (32.1)
Master's degree	20 (23.8)
Doctoral degree	6 (7.1)
Family income in US\$	
<15 000	2 (2.4)
15-24 999	2 (2.4)
25-34 999	6 (7.1)
35-49 999	8 (9.5)
50-74 999	13 (15.5)
75-99 999	13 (15.5)
>100 000	39 (46.4)
No response	1 (1.2)
Feeding method pre-frenotomy	
Exclusively breastfed at the breast	30 (35.7)
Fed with a bottle only	13 (15.5)
Both breastfed at the breast and fed with a bottle	41 (49.8)
Feeding method post-frenotomy	
Exclusively breastfed at the breast	40 (47.6)
Fed with a bottle only	10 (12)
Both breastfed at the breast and fed with a bottle	34 (40.4)

Abbreviation: GED, General Educational Development.

Infants with more severe tongue-tie demonstrated greater improvement in GIGER scores post-frenotomy ( $B = 14.327, t = 1.983$ ), but this was not statistically significant ( $P = .051$ ). Infants with more severe tongue-tie had greater improvements in the Common GI and GER Symptoms subscale ( $B = 10.386, t = 2.245, P = .02$ ). Younger infants had greater improvement in GIGER scores after tongue-tie correction ( $B = -0.716, t = -2.008, P = .04$ ).

**Table 3.** Demographics of Participants' Infants (N = 84).

Characteristic	No. (%)
Infant age	
<2 months	64
2-<4 months	13
4-<7 months	7
Infant sex	
Male	47
Female	37
Infant race	
American Indian/Alaskan Native	1
Asian	1
White	72
More than 1 race	8
Other	2
Infant ethnicity	
Hispanic/Latino	3
Not Hispanic/Latino	73
Other	5
Unknown	2
Not answered	1
Infant medical diagnoses	
GERD	10
Hearing impairment	1
Cardiac disorder	3
Umbilical hernia	3
Developmental delay	1
Laryngomalacia	1
Tongue-tie severity	
II	1
III	75
IV	8

Abbreviation: GERD, gastroesophageal reflux disease.

Parents reported up to 9 different reflux strategies prior to frenotomy, and 7 post-frenotomy. The most common strategies used prior to tongue-tie correction were holding the infant upright after feeding ( $n = 9$ ), paced feedings ( $n = 6$ ), and use of reflux medications ( $n = 6$ ). Following the procedure, the most common strategies were holding infant upright after feeding ( $n = 12$ ), elevating the head of the bed ( $n = 7$ ) and paced feedings ( $n = 7$ ). Six parents reported use of reflux medications prior to tongue-tie correction, while only 1 reported using pharmacotherapy to treat reflux post-frenotomy. However, this difference was not statistically significant ( $P = .09$ ). None of the nonpharmacologic strategies were significantly different pre-frenotomy to post-frenotomy despite parental perception of infant reflux symptoms being significantly lower post-frenotomy ( $B = -1.239, P < .01$ ). Total strategies used were not significantly different at the 2 time points.

**Table 4.** Difference in GIGER Scores Pre-Frenotomy and Post-Frenotomy.

GIGER scale	Pre-frenotomy	Post-frenotomy	z	P
Total score	51.88	37.94	-5.204	<.001
Difficulties with self-regulation	12.79	10.02	-3.766	<.001
Common symptoms	31.17	22.54	-4.960	<.001
Compelling symptoms	7.93	5.38	-3.982	<.001

Abbreviation: GIGER, gastrointestinal and gastroesophageal reflux.

## Discussion

This is the first study to utilize the GIGER Scale, a comprehensive measure of GI and GER symptoms, to compare symptomology before and after tongue-tie correction in infants below the age of 7 months. Total and subscale scores on the GIGER were significantly lower post-frenotomy, suggesting a relationship between these symptoms and tongue-tie. This finding was also clinically significant, supported by parents reporting fewer subjective symptoms in their infants 2 weeks following tongue-tie correction. There were no significant differences in overall strategies used to manage reflux from pre-frenotomy to post-frenotomy, so the change in symptoms was not a result of increased management strategies. In fact, 5 of the 6 infants who were prescribed reflux medications were no longer taking these at the time of follow-up. Given the increasing evidence of adverse effects from acid-reducing medications,<sup>22-25</sup> this is a clinically important finding even though it was not statistically significant.

The underlying mechanisms for why frenotomy would cause a reduction in symptoms of GI and GER are not well understood. Alterations in tongue mechanics due to tongue-tie may affect the infant's ability to effectively latch onto the breast or bottle, causing the infant to swallow excessive air (ie, aerophagia).<sup>7</sup> While it has not been studied to our knowledge, swallowing excessive air may lead to increased volume in the stomach, placing added pressure on the lower esophageal sphincter and potentially placing the infant at risk for symptoms of GER.<sup>26</sup> This pathophysiology may explain GER symptoms, but would not explain improvement in symptoms along the lower GI tract.

Possible explanations for changes in lower GI symptomatology may include changes in feeding method, type of milk, or changes in the composition of the milk due to changes in the latch at breast. In this sample, 10 of the infants (8.4%) were transitioned to exclusive breastfeeding post-frenotomy. An increase in the amount of human milk consumed or the amount of direct feeding at breast may contribute to improvement in GI and GER symptoms. Human milk contains enzymes that aid in digestion and are more easily digested than formula.<sup>27</sup>

In addition, feeding directly at breast has been found to alter the microbiota of the infant<sup>28</sup> by exposing infants to the bacteria of the mother's breast. Changes in the mechanics of breastfeeding after frenotomy in infants with tongue-tie may also contribute to improvement in GI and GER symptoms. Prior to frenotomy, infants with tongue-tie may only achieve a shallow latch. It is possible that this altered latch, or short feedings at the breast as the infant tires from the difficulty latching, may result in the infant only obtaining the foremilk from the breast, which is compositionally different from milk at the end of the feeding (ie, hindmilk).<sup>29</sup> If frenotomy allows infants with tongue-tie who are breastfeeding to achieve a better and deeper latch, there may be changes in the composition of the milk that they are able to receive at breast, which could also contribute to symptom improvement.

Tongue-tie severity, as measured by Kotlow diagnostic criteria, was only found to have an effect on 1 subscale of the GIGER post-frenotomy, with infants with more severe tongue-tie having greater improvement in symptoms on the common GI and GER symptoms subscale. While we would expect that infants with more severe tongue-tie would experience greater improvement than those with less severe tongue-tie, the lack of significant findings may indicate that anatomical classification alone is not as important as the function of the tongue. Infants had improvement in GI and GER symptoms regardless of degree of severity of their tongue-tie.

Finally, younger infants were found to have greater improvement in GI and GER symptoms post-frenotomy. This may be related to prolonged GI and GER disturbance in older infants, which could take longer to resolve,<sup>30</sup> leading to less improvement in symptoms by the 2-week follow-up in this study. Future research is necessary to understand how the GI mucosa, gastric pH, or the microbiome of the GI tract responds to frenotomy intervention respective to infant age at the time of treatment. These data may provide guidance for optimal age to treat tongue-tie.

The main limitation of this study was that the sample was homogeneous. Most infants were diagnosed with

severe tongue-tie and all were treated for concurrent lip-tie. Future research should aim for greater diversity in the sample, including greater variation of tongue-tie severity and comparison of GER and GI symptoms in infants with and without lip-tie. At present, lip-tie has been inconsistently defined in the literature,<sup>31</sup> and we do not have research to explain the role of lip-tie in breastfeeding difficulties or how an upper lip restriction may create GI or GER symptoms.<sup>31,32</sup> However, it has been noted that upper lip ties may impair seal onto the breast, leading to aerophagia.<sup>6,33</sup> In addition, there were some parents (n = 18) who completed the first survey but did not complete the second survey and thus were not included in these analyses. Respondents who completed the follow-up survey were less likely to have prior experience with breastfeeding ( $t = 2.063$ ,  $P = .04$ ) compared with nonrespondents. This difference is not thought to have contributed to results of GI and GER symptoms recorded through parental report.

This study did not collect longitudinal data beyond the 2-week follow-up; it would be helpful to reevaluate infant symptoms over a longer time span to determine whether and how GI and GER symptoms change over time. Despite these limitations, these findings have made an important contribution to our understanding of the impact of tongue-tie and its correction on GER and GI symptoms in young infants and support the need for further research.

Parental concerns with feeding and infant discomfort with eating are a significant source of stress for new parents. The results of this study suggest that earlier identification of symptoms and infants with more severe tongue-tie will achieve the greatest improvement in GI tract symptomatology. Our findings support prior work that demonstrated improvement in GI and GER symptoms following frenotomy. In addition, these findings provide more comprehensive data on how the GI tract may be affected by this oral anomaly, with clear directions for future research. For infants who display symptoms suggestive of GI or GER dysfunction, or necessitate acid-reducing medication, clinicians should consider tongue-tie as a differential diagnosis causing these symptoms. Providers can utilize and interpret the GIGER scale to ascertain symptoms that may indicate tongue-tie necessitating feeding support or advocacy for specialty referral.

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### Author Contributions

RRH: Contributed to the design of the work, acquisition, analysis, and interpretation of data; drafted and revised the

intellectual content; gave final approval of manuscript submitted; agrees to be accountable for all aspects of the work.

BFP: Contributed to the design of the work, analysis, and interpretation of data, drafted and revised the intellectual content, gave final approval of manuscript submitted; agrees to be accountable for all aspects of the work.

### Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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### Ethical Approval

This study was approved by Boston College Institutional Review Board (IRB no. 20.267.01).

### Participant Consent

All participants consented to participate in the study using an online consent form.

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