

## REVIEW ARTICLE

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

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## Abstract

**Aim:** The aims of this systematic review were to first identify and summarise original research that compared symptoms of problematic feeding in infants with tongue tie before and after frenotomy and then evaluate the quality of measures used to assess problematic feeding.

**Methods:** CINAHL and PubMed were searched for ((tongue-tie) or (ankyloglossia) and ((feeding) or (breastfeeding) or (bottle-feeding)) and ((frenotomy) or (frenectomy) or (frenulectomy) or (frenulotomy)). Original research reporting on feeding before and after frenotomy in infants under 1 year old was included.

**Results:** Maternal nipple pain, breastfeeding self-efficacy and LATCH scores improved after frenotomy. Few data are available on the effect of frenotomy on infant feeding. The measures used to assess infant feeding were not comprehensive and did not possess strong psychometric properties.

**Conclusion:** Literature suggests that maternal nipple pain, self-efficacy and LATCH scores improve in breastfeeding mother-infant dyads after frenotomy. However, current literature does not provide adequate data regarding the effect of frenotomy on the infant's ability to feed or which infants benefit from the procedure. Future research should utilise comprehensive, psychometrically sound measures to assess infants for tongue tie and to evaluate infant feeding to provide stronger evidence for the effect of frenotomy on feeding in infants with tongue tie.

## KEYWORDS

ankyloglossia, bottle feeding, breastfeeding, bottle feeding, feeding behaviour, frenotomy

## 1 | INTRODUCTION

Ankyloglossia, commonly referred to as tongue tie, is a congenital anomaly where the tongue is abnormally tightly connected to the

floor of the mouth, limiting its mobility.<sup>1</sup> This anomaly has been cited in literature dating back to the 1700s<sup>2</sup> and was routinely treated up through the 1940s, when infant formula use increased and breastfeeding rates declined.<sup>3</sup> Prevalence of tongue tie has been reported

**Abbreviations:** BBAT, bristol breastfeeding assessment tool; BSES, breastfeeding self-efficacy scale; BSES-SF, breastfeeding self-efficacy scale-short form; CINAHL, cumulative index of nursing and allied health literature; EBF, exclusive breastfeeding; FDRBI, frenotomy decision rule for breastfeeding infants; FDTBD, frenotomy decision tool for breastfeeding dyads; GERD, gastroesophageal reflux disease; HATLFF, Hazelbaker Assessment Tool for Lingual Frenulum Function; IBFAT, infant breastfeeding assessment tool; I-GERQ, infant gastroesophageal reflux questionnaire; I-GERQ-R, infant gastroesophageal reflux questionnaire-revised; LATCH, latch, audible swallowing, type of nipple, comfort, hold; NRS, numeric rating scale; PRISMA, preferred reporting items of systematic reviews and meta-analyses; RCT, randomised control trial; SF-MPQ, short-form McGill pain questionnaire; VAS, visual analogue scale.

as low as 0.3%<sup>4</sup> to as high as 16%,<sup>5</sup> with a hereditary component and male predominance.<sup>6</sup>

Organisations such as the Centers for Disease Control and the World Health Organization provide recommendations for exclusive breastfeeding (EBF) for the first 6 months of life.<sup>7</sup> Healthy People 2020 sought to increase exclusive breastfeeding rates to 25.5%.<sup>8</sup> As of 2016, the exclusive breastfeeding rate at 6 months in the United States was 25.4%.<sup>9</sup> Exclusive breastfeeding through 6 months remains an objective in the preliminary development of Healthy People 2030, signifying that additional work is needed to increase this rate for more mother-infant dyads to obtain the benefits associated with breastfeeding.<sup>10</sup> A greater understanding of why mothers stop breastfeeding sooner than planned may help increase breastfeeding rates. The reasons for breastfeeding cessation are complex and multidimensional. Socioeconomic factors, employment status, competing family demands, food insecurity, breastfeeding discomfort and concerns surrounding infant weight gain and nutrition are all contributing factors that lead to early breastfeeding cessation.<sup>11-13</sup>

There are many factors that lead to early cessation of EBF, with one of the most commonly reported reasons being nipple pain.<sup>14</sup> Tongue tie has been associated with nipple pain,<sup>15</sup> and frenotomy to treat tongue tie is thought to reduce breastfeeding issues. A previous analysis of existing literature in 2017 identified studies with small sample sizes, few RCTs, a lack of longitudinal data and limited evidence on the effects on infant feeding, with the most notable improvement in maternal reports of nipple pain.<sup>16</sup> Other reviews on tongue tie have assessed trends in treatment and the use of frenotomy for tongue tie in breastfed infants.<sup>17-19</sup> None of the previous reviews critiqued the quality of the measures used to evaluate infant feeding. Assessment of the quality of an infant feeding measure includes reviewing the psychometric properties of the measure, including reliability and validity, and reviewing whether the measure comprehensively evaluates the problem being studied (ie the infant's feeding skills and behaviours). Evidence of validity supports that the measure assesses the infant's feeding, and not a related, but different construct, such as strategies used to support feeding. Evidence of reliability supports that the score is an accurate reflection of the truth and that the measure performs in a consistent way across time or across people conducting the assessment. Research studies that fail to use outcome measures with evidence of strong psychometric properties may not accurately measure the problem being investigated.<sup>20</sup>

Despite the low strength of the evidence, a recent study reported an 866% increase in frenotomy rates from 1997 to 2012.<sup>21</sup> The impact of frenotomy on infant feeding difficulties needs to be studied. The purpose of this systematic review was twofold. First, original research that compared symptoms of problematic feeding before and after frenotomy in infants under 1 year old with tongue tie was reviewed. Next, the quality of the feeding-related outcome measures and psychometric properties of the assessment tools used were reviewed to develop a better understanding of the quality of measures used to assess symptoms of problematic feeding in each study. Through these two aims, the strengths and limitations of the

## Key notes

- The current literature supports frenotomy to reduce short-term maternal nipple pain in breastfeeding mothers.
- The evidence to date has not utilised comprehensive measures of infant feeding to evaluate the effects of frenotomy for the infant with tongue tie.
- Future research that focuses on physiologic changes in the infant post-frenotomy is needed to support the use of frenotomy for the treatment of tongue tie.

research and assessment tools are explicated, and implications for practice and research are presented.

## 2 | METHODS

### 2.1 | Search method

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

### 2.2 | Study selection

Articles that compared infant feeding from the maternal and/or infant perspective both before and after treatment of tongue tie using frenotomy were examined for inclusion in this review. Articles were excluded if they were not original research, if it was a case report on fewer than three children, child age at the time of treatment was over 12 months, or if data were not collected both prior to and following frenotomy. Additionally, studies were excluded if they were found to not measure feeding or if the sample only included infants with a different anatomical anomaly (not tongue tie). Titles and abstracts were screened independently by both authors using the inclusion and exclusion criteria, and discrepancies were resolved. Articles that appeared to meet inclusion and exclusion criteria or that could not be properly evaluated based on the abstract were reviewed in full text. Both authors then reviewed the full-text articles for inclusion/exclusion independently. If one author omitted or selected an article, both authors reviewed the full text together to discuss and a final decision

was made to include or exclude the article based on the aims of this review. The reference lists of these papers were screened for any additional articles; three articles were found in reference lists that were included in the review.

## 2.3 | Study characteristics extracted

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

## 2.4 | Assessment of psychometric properties of outcome measures

After the feeding-related outcome measures were identified from the studies that met inclusion criteria, a review of the literature was conducted for each of these assessment tools to identify the psychometric properties. A search was conducted in both PubMed and

CINAHL using the full name of the assessment tool. The search was limited to humans, English language, full text and with children aged birth to 12 months. Articles reporting on the psychometric properties of the feeding-related measures were used to evaluate the quality of assessments used for studying the effect of frenotomy on feeding to date.

## 3 | RESULTS

### 3.1 | Study selection

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

### 3.2 | Study characteristics

#### 3.2.1 | Sample size & characteristics

The median sample size of the studies was 58 (range 14-246). Four of the studies had 30 or fewer infants, while seven studies included more than 100 infants. Most of the studies recruited mother-baby dyads that presented with breastfeeding difficulty despite lactation

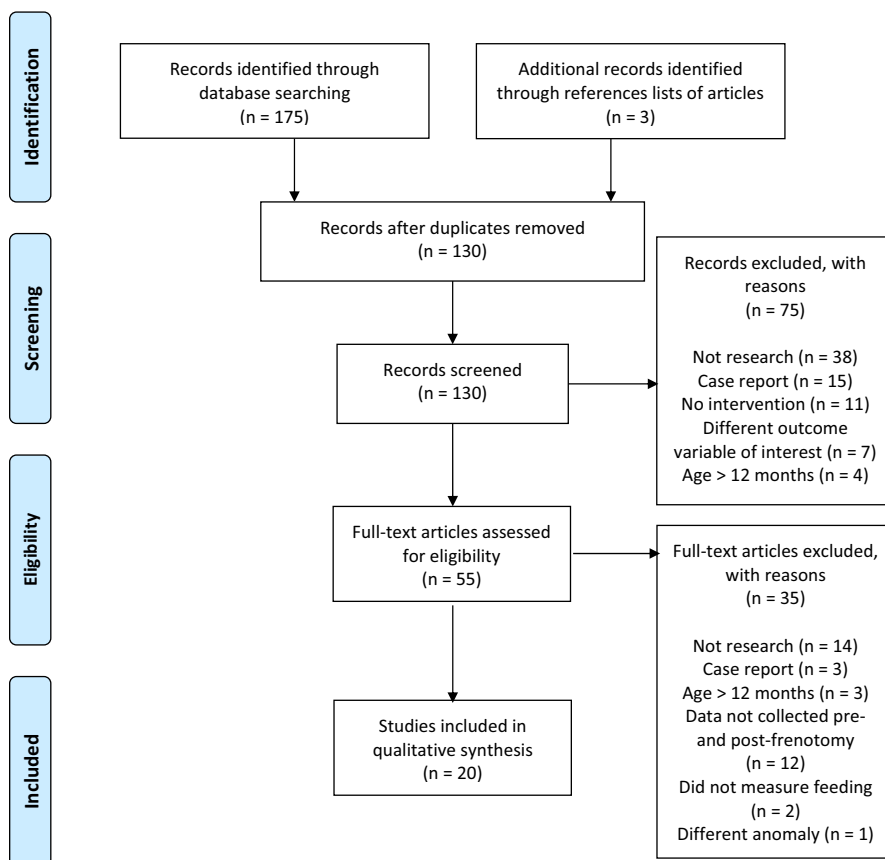


FIGURE 1 PRISMA 2009 flow diagram. From: Moher et al<sup>69</sup>

**TABLE 1** Characteristics of included studies (N = 20)

Author/Year	Sample characteristics	Assessment/treatment	Outcomes measured	Outcome time points
Ballard, Auer, & Khoury (2002) <sup>22</sup>	N = 123 93 male, 61 female	Hazelbaker/Scissors	Nipple Pain (VAS) Subjective Maternal Symptoms	Immediately pre- and post-frenotomy
Braccio et al (2016) <sup>23</sup>	N = 158 Median age at frenotomy 2 wk (range 1 d-5 mo)	Not stated/Not stated	Maternal report of feeding problems	Immediately pre-frenotomy, 48 h post-frenotomy and additional follow-up around 4-wk post-frenotomy
Buryk, Bloom & Shope (2011) <sup>31</sup>	N = 58 Randomly assigned to frenotomy (30) or usual care (28), Mean age 6 d (all ≤ 30 d old) 38 males, 20 females	Hazelbaker/Not stated	Nipple pain (SF-MPQ), IBFAT	Immediately pre- and post-frenotomy and 2-wk follow-up
Dollberg, Marom & Botzer (2014) <sup>32</sup>	N = 244 Median age 14 d (range 1-135 d) 143 male, 101 female Nipple pain and latch difficulty led to referral	Coryllos/Not stated	Maternal report of feeding problems	Pre-frenotomy, 2 wk, 3-mo and 6-mo post-frenotomy
Emond et al (2014) <sup>41</sup>	N = 107 Randomly assigned to frenotomy (55) or usual care (52) Mean age 62 d Excluded if Hazelbaker score < 6 (severe TT)	Hazelbaker/Not stated	LATCH, IBFAT, BSES-SF, Nipple pain (VAS)	Pre-frenotomy, 5-d and 8-wk post-frenotomy
Geddes et al (2008) <sup>33</sup>	N = 24 Mean age 33 d (±28 d, range 4-131 d)	Not stated/Scissors	24-h milk production, LATCH, nipple pain (VAS), tongue movement via ultrasound	Immediately pre-frenotomy and within 7-d post-frenotomy
Ghaehri et al (2017) <sup>34</sup>	N = 237 Mean age 4.4 wk 86% White 133 males, 104 females	Kotlow and Coryllos/Diode laser	BSES-SF, I-GERQ-R, nipple pain (VAS)	Immediately pre-frenotomy, 1-wk and 1-mo post-frenotomy
Ghaehri, Cole & Mace (2018) <sup>35</sup>	N = 54 Mean age 8.3 wk (range 7 d-37 wk) 43 white 28 male, 26 female	Kotlow and Coryllos/Diode Laser	BSES-SF, I-GERQ-R, nipple pain (VAS)	Immediately pre-frenotomy, 1-wk and 1-mo post-frenotomy
Hogan, Westcott & Griffiths (2005) <sup>39</sup>	N = 57, mean age 19 d (range 3-70) 14 males, 14 females in treatment group 1.3:1 male to female in control group	Not stated/Scissors	Maternal report of feeding difficulties	Immediately pre-frenotomy and 24 h, weekly × 4 wk and at 4 mo Frenotomy group (n = 28) compared with control group (n = 29)
Illing et al (2019) <sup>36</sup>	N = 176 Mean age 44 d 109 males, 67 females	Kotlow/Scissors	Maternal report of feeding problems	Immediately pre-frenotomy and approximately 3-wk post-frenotomy (mean time 23 d)
Khoo et al (2009) <sup>37</sup>	N = 62 40 males, 20 females Mean age 23.5 d (± 17.1)	Not stated/Scissors	Maternal reports of feeding symptoms and degree of difficulty	Immediately pre-frenotomy and 3-mo follow-up

(Continues)

TABLE 1 (Continued)

Author/Year	Sample characteristics	Assessment/treatment	Outcomes measured	Outcome time points
Martinelli et al (2015) <sup>40</sup>	N = 28 20 male, 8 female Frenotomy at 45 d of age	Not stated/Scissors	Maternal report of feeding problems, number of sucks and length of pause between sucks	Day 30 of life (pre-frenotomy) and day 75 of life post-frenotomy, compared between frenotomy and control group (N = 14 each)
Miranda & Milroy (2010) <sup>38</sup>	N = 51 completed study Age range 12-36 d	Not stated/Scissors	Infant weight, number of feeding sessions/24 h, nipple pain (NRS)	Immediately pre-frenotomy and 2-wk follow-up
Muldoon et al (2017) <sup>24</sup>	N = 89 Mean age at frenotomy 7 wk 3 d	Not stated/Varied	Nipple pain (VAS), modified LATCH	Immediately pre-frenotomy and 1-mo post-frenotomy
Schlatter et al (2019) <sup>25</sup>	N = 30 Characteristics of sample undergoing frenotomy not described	Hazelbaker/Not stated	LATCH, BBAT, maternal report of feeding problems	Immediately pre-frenotomy and 2.5-wk post-frenotomy
Sethi et al (2013) <sup>26</sup>	N = 52 Mean age 19 d (range 3-120) 35 male, 17 female All mothers had BF problems prior to referral	Not stated/Scissors	Maternal report of feeding symptoms, rate of EBF post-frenotomy	Pre-frenotomy and within 5-mo post-frenotomy
Sharma & Jayaraj (2015) <sup>42</sup>	N = 42 36 had frenotomy 23 males, 19 females Median age at treatment 38 d (range 15-178 d)	Not stated/Scissors	IBFAT	Immediately pre-frenotomy and 1-mo post-frenotomy, compared with those who did not have frenotomy (n = 6)
Srinivasan, Dobrich, Mitnick, & Feldman (2006) <sup>27</sup>	N = 27 18 male, 9 female Mean age 19 d (range 2-71)	Frenotomy Decision Rule for Breastfeeding Infants/Scissors	LATCH, nipple pain (SF-MPQ), maternal feeding questionnaire	Immediately pre- and post-frenotomy and 3-mo telephone survey
Srinivasan et al (2019) <sup>28</sup>	N = 30 Mean age 37.9 d (range 9-80) 20 male, 10 female	Coryllos and Frenotomy Decision Tool for Breastfeeding Dyads/Scissors	LATCH, nipple pain (VAS), maternal report of improvement in BF	Immediately pre- and post-frenotomy and 2-, 7- and 14-d post-frenotomy
Wakhanrittee, Khorana & Kiatipunsodsai (2016) <sup>30</sup>	N = 246 147 male, 99 female Median age at frenotomy 50 h 142 had severe TT 180 had moderate TT	Mild; Moderate; Severe/Scissors	LATCH, nipple pain (NRS), infant feeding pattern, EBF rate at follow-up	Immediately pre-frenotomy, 24-h, 1-wk and 3-mo post-frenotomy

Abbreviations: BBAT, Bristol breastfeeding assessment tool; BF, Breastfeeding; EBF, Exclusive breastfeeding; IBFAT, Infant breastfeeding assessment tool; LATCH, Latch, Audible swallowing, Type of nipple, Comfort, Hold; NRS, Numeric rating scale; SF-MPQ, Short-form McGill pain questionnaire; VAS, Visual analogue scale.

support,<sup>22-38</sup> and four of the studies screened all infants born during the study period.<sup>22,25,39,40</sup> Not all studies listed explicit inclusion or exclusion criteria. Beyond breastfeeding difficulties, some studies restricted inclusion to certain infant age ranges,<sup>27,28,34-37,41</sup> specific tongue tie severity<sup>22,31,41</sup> or baseline LATCH score.<sup>41</sup> Exclusion criteria included the presence of other oral anomalies,<sup>25,28,32,40,41</sup> prematurity,<sup>25,33-35,41</sup> medical co-morbidities,<sup>25,34,35,40</sup> maternal breast surgery<sup>34,35</sup> and delivery complications.<sup>40</sup>

There was variety in maternal and infant demographic information reported across studies. The most commonly reported information was infant age, sex and birth weight. One study did not report infant age,<sup>22</sup> and sex was reported in 14 of the studies.<sup>22,26-28,30-32,34-37,39,40,42</sup> Birth weight was reported in four studies,<sup>28,30,37,38</sup> and current infant weight was reported in one.<sup>30</sup> Only three reported infant ethnicities. In all three, the majority of infants were white.<sup>34-36</sup> One study discussed how maternal

ethnicity, education level and socioeconomic status differed between the overall population at the hospital and those that enrolled in the study but did not report the actual data comparing the sample to the overall population.<sup>41</sup> None of the other 19 studies reported maternal ethnicity, socioeconomic status or education level. Maternal data collected included age,<sup>27,28,30,36</sup> parity<sup>24,28,32</sup> and medical history.<sup>27,28</sup> Family history of tongue tie was reported in three studies.<sup>24,25,37</sup> Of the 20 studies, four were RCTs,<sup>31,32,39,41</sup> all of which had small sample sizes. The remaining studies obtained participants through convenience sampling, a form of selection bias. Additional risks of bias (eg performance, confounding, channelling, validity) are discussed in the details of the studies described below.

### 3.2.2 | Ankyloglossia assessment tools used

Across the 20 studies, tongue tie was diagnosed using differing methods. Four studies used the Hazelbaker Assessment Tool for Lingual Frenulum Function<sup>43</sup> (HATLFF),<sup>22,25,31,41</sup> one used Kotlow's diagnostic criteria<sup>36,44</sup> and one used the Coryllos Grading System.<sup>32,45</sup> Srinivasan used the Frenotomy Decision Rule for Breastfeeding Infants (FDRBI) in the 2006 publication<sup>27</sup> and then used a revised version of this tool, called the Frenotomy Decision Tool for Breastfeeding Dyads (FDTBD), in their 2019 publication.<sup>28</sup> One study classified tongue tie as mild, moderate or severe based on their own definition without evidence for the definition.<sup>30</sup> Three of the studies used a combination of two different assessment tools (eg Kotlow's and Coryllos).<sup>28,34,35</sup> Nine of the studies did not report if a standardised diagnostic assessment measure was used.<sup>23,24,26,33,37-40,42</sup>

The diagnostic tools used assess different characteristics of tongue tie. The HATLFF focuses on tongue movement, while the Coryllos Grading System classifies tongue tie based on severity of restriction to the floor of the mouth. Kotlow's diagnostic criteria involves measurement of free tongue (portion not attached to the frenulum) in millimetres. The FDRBI evaluates both the mother and infant. The mother is assessed for pain/trauma with breastfeeding, and infant is assessed for his/her ability to sustain a latch onto the breast and weight gain. The infant's tongue is examined for the presence of an anterior frenulum and tongue movement specific to elevation, cupping and protrusion. There is no score requirement for the FDRBI.<sup>27</sup> The newer version, the FDTBD, evaluates these same characteristics in mother and baby. In addition, feeding time and milk transfer are subjectively reported by the mother. Lateral tongue movement is assessed along with elevation, suction and protrusion. The maternal and infant categories are scored and if one or more symptoms are present in both the mother and infant (score of two or more), frenotomy is recommended.<sup>28</sup>

To date, very little has been published on the psychometric properties of the assessment tools used to diagnose tongue tie. The Kotlow, Coryllos, FDTBD and FDRBI assessment tools have no published psychometrics. The HATLFF has demonstrated interrater reliability in the measurement of tongue mobility (elevation, extension

and lateralisation) when comparing infants with and without tongue tie.<sup>39</sup> However, researchers found that the HATLFF resulted in a large number of unclassified infants, potentially missing tongue tie diagnosis,<sup>46</sup> as well as false negatives.<sup>47</sup> Others acknowledged subjectivity of the HATLFF between raters.<sup>41</sup>

### 3.2.3 | Frenotomy methods

Twelve of the studies included in this review used scissors as the method for frenotomy,<sup>22,26-28,30,33,36-40,42</sup> two used laser,<sup>34,35</sup> one reported "varied" methods,<sup>24</sup> and five studies did not indicate the method of treatment.<sup>23,25,31,32,41</sup> There were no adverse effects associated with frenotomy in any of the studies.

### 3.2.4 | Feeding-related outcomes

To address the first aim of this paper, we reviewed feeding-related outcomes, which include symptoms of feeding difficulty, evaluation of breastfeeding and maternal report of satisfaction with breastfeeding. The primary feeding-related outcomes measured included nipple pain, breastfeeding assessment, breastfeeding self-efficacy and infant gastroesophageal reflux. Eleven of the 20 studies utilised subjective assessment of improvement in breastfeeding via maternal report.<sup>22,23,25-28,32,36,37,39,40</sup> All eleven reported subjective improvements in breastfeeding, including decreased nipple pain, improvements in latch and shorter feeding times. Subjective nipple pain improved immediately after frenotomy was performed. Dollberg<sup>32</sup> stated 90% of breastfeeding problems were alleviated after frenotomy; however, three percent of women reported worsening of symptoms at 2-week post-frenotomy.

One study included breastfed and artificially fed infants. This study measured subjective outcomes on bottle feeding pre- and post-frenotomy, including total feeding time, amount of dribbling from the bottle nipple and maternal report of infant gassiness.<sup>39</sup> Subjective improvements in feeding time, dribbling and gas were noted by the mothers of the eight infants in the treatment group with no improvements in the nine babies in the control group ( $P < .001$ ).<sup>39</sup> Wakhanrittee found that frenotomy performed 24 hours or less after birth ( $P < .001$ ), less severe tongue tie ( $P < .001$ ), female sex ( $P = .01$ ) and more children in the family ( $P = .02$ ) significantly contributed to EBF success at 3-month post-frenotomy.<sup>30</sup> Sharma et al<sup>42</sup> demonstrated similar findings, with mothers of infants less than 30 days old having greater improvements in breastfeeding symptoms, although the difference was not significant ( $P = .09$ ). An increased number of sucks and a decrease in pause length between suck groups was noted in infants post-frenotomy when compared to infants that did not undergo the procedure. Geddes found that breastmilk production was significantly increased in the right breast, with changes in feeding mechanics on ultrasound after frenotomy. In this study, only six of the 24 mothers completed post-frenotomy milk transfer measurements. Pre-frenotomy feeding mechanics revealed biting on the

nipple or an inability to maintain an effective seal, both of which cause nipple pain.<sup>33</sup>

### 3.2.5 | Nipple pain

The most frequent outcome measured was nipple pain, evaluated in 19 of the 20 studies, and the one study that did not measure improvements in nipple pain mentioned it as the most common presenting symptom.<sup>42</sup> Assessment of nipple pain was completed pre- and post-frenotomy in all 19 studies, using a variety of assessment techniques. Pain rating using a visual analogue scale (VAS)<sup>22,24,34,35,41</sup> or subjective maternal report<sup>23,25,26,32,36,37,39,40</sup> were the most commonly used methods to assess nipple pain. A numeric rating scale (NRS) was used in four studies,<sup>28,30,33,38</sup> and the Short-Form McGill Pain Questionnaire (SF-MPQ) was used in two.<sup>27,31</sup> In all 19 studies, there was a decrease in maternal nipple pain post-frenotomy, with improvements occurring immediately post-frenotomy and up through 2-week post-procedure. The severity of tongue tie was not correlated with the degree of pain or improvement in maternal symptoms post-procedure.<sup>37,39</sup> Khoo et al<sup>37</sup> found that mothers who presented with nipple pain as the main symptom of tongue tie were more likely to be breastfeeding 3-months post-frenotomy (OR 5.8, 95% CI 1.1-31.6).

### 3.2.6 | Breastfeeding assessment

In addition to the studies that discussed subjective reports of breastfeeding, nine studies reported on breastfeeding assessment using a standardised measure. Three of the studies utilised the Infant Breastfeeding Assessment Tool (IBFAT) as a measure of frenotomy success.<sup>31,41,42</sup> Buryk, Bloom & Shope<sup>31</sup> conducted a single-blinded RCT, measuring scores on the IBFAT immediately before and after frenotomy and again at 2-week post-procedure. Infants were randomly assigned to the frenotomy or control groups, with no statistically significant differences between groups prior to frenotomy ( $P = .44$ ). The IBFAT scores improved significantly ( $P = .03$ ) in the frenotomy group immediately after the procedure. Two-week comparison of IBFAT scores was not possible, as frenotomies were performed in all but one infant assigned to the control group. It is important to note that the version of the IBFAT used in this study consisted of four categories with a total possible score of 15, which is not consistent with the original tool development, that allows for a maximum score of 12.<sup>48</sup>

Emond et al<sup>41</sup> conducted a randomised parallel group trial, evaluating breastfeeding at the time of tongue tie diagnosis, 5 days after frenotomy and again at 8 weeks post-procedure. A control group was assigned, with 44 of 52 infants undergoing frenotomy before the 8-week follow-up. This RCT did not show any statistically significant improvements in breastfeeding via the IBFAT ( $P = .76$ ). Sharma & Jayaraj<sup>42</sup> conducted IBFAT scoring via telephone survey pre-frenotomy and 1-month post-frenotomy. A statistically

significant improvement in breastfeeding was noted by mothers in the frenotomy group, compared with those whose infants did not have the frenotomy procedure ( $P < .001$ ). The severity of tongue tie was not consistently documented across studies, and long-term effects were not assessed. The study by Emond et al<sup>41</sup> evaluated tongue tie severity using the HATLFF; however, only those with mild-moderate tongue tie were included, limiting generalisability to that subset of the population.

Six studies utilised the LATCH tool,<sup>25,27,28,30,33,41</sup> with a seventh adopting a modified version of the LATCH that has not been validated.<sup>24</sup> Srinivasan et al<sup>27</sup> assessed breastfeeding using the LATCH tool immediately before and after frenotomy, with a statistically significant improvement in LATCH scores following frenotomy ( $P < .001$ ). There were no differences in LATCH scores in the Emond<sup>41</sup> study between groups. Another study by Srinivasan et al<sup>28</sup> in 2019 assessed breastfeeding using the LATCH tool pre-frenotomy, immediately post-frenotomy and again at days 2, 7 and 14 post-frenotomy. This study demonstrated statistically significant increases in LATCH scores immediately post-frenotomy and again from days seven to 14 post-frenotomy ( $P < .001$ ). Samples sizes were highly variable in these studies, ranging from 26 to 246, and lack longitudinal data beyond 8 weeks post-revision. The studies by Srinivasan, Dobrich, Mitnick & Feldman (2006) and Geddes et al (2008) did not use a validated tool to diagnose tongue tie, had small sample sizes ( $N = 30$  and  $24$ , respectively) and lacked control groups.<sup>27,33</sup> As previously mentioned, while Emond (2014) was an RCT, the authors limited the sample to those with mild-moderate tongue tie and infants less than 2-weeks of age. Muldoon et al reported a significant improvement in LATCH scores post-frenotomy (MD  $-0.50$ , 95% CI  $-0.67$  to  $-0.33$ ), using a modified version of the tool. While breastfeeding improved and maternal pain decreased, the rate of formula use doubled at 1-month post-frenotomy.<sup>24</sup>

Srinivasan<sup>28</sup> utilised frenotomy alongside lactation counselling, with a small sample size ( $N = 30$ ), with several participants lost to follow-up. It is not possible to determine the independent influences of frenotomy on outcomes as concurrent lactation support was also included. The study by Wakharrittee et al had the largest sample, consisting of 246 infants diagnosed with tongue tie. LATCH scores were significantly increased at 24-hours and 1-week post-frenotomy ( $P < .001$ ). However, this study also lacked a control group and endorsed a possible selection bias of motivated mothers eager to breastfeed.<sup>30</sup> Significant improvements in LATCH scores were also noted by Schlatter at 2.5-weeks post-frenotomy ( $P = .01$ ). This same study also used the Bristol Breastfeeding Assessment Tool (BBAT) and demonstrated a significant improvement in the BF assessment post-frenotomy ( $P = .01$ ), with results limited by a small sample size ( $N = 23$ ) and loss of participants to follow-up.<sup>25</sup>

### 3.2.7 | Breastfeeding self-efficacy

The 14-item Breastfeeding Self-Efficacy-Short Form (BSES-SF) was used in three of the 20 studies included in this systematic

TABLE 2 Psychometric properties of assessment tools

Tool (Study)	Purpose and intended user	Target population	Items: number and constructs	Reliability	Validity
Bristol Breastfeeding Assessment Tool (BBAT) (Ingram et al, 2015 <sup>44</sup> )	Rapid breastfeeding assessment Provider or Parent	Healthy full-term infants Tested in infants up to 10 wk old	4 items: Positioning, Attachment, Sucking and Swallowing	Interrater ICC = 0.78 Cronbach's alpha = 0.67	Construct validity with breastfeeding self-efficacy scale ( $r = 0.57$ )
Infant Breastfeeding Assessment Tool (IBFAT) (Matthews, 1988 <sup>48</sup> )	Assess infant competence with breastfeeding Provider or Parent	Healthy, full-term infants	6 items: Signalling, Rooting, Fixing and Sucking Pattern	77%-91% interrater agreement Reliability 0.27-0.78	Does not support predictive validity or discriminate validity Moderate convergent validity ( $r_s = 0.56-0.63$ ) Construct validity $r = 0.69$ with LATCH, 0.78-0.86 with MBA
LATCH (Jensen et al 1994 <sup>58</sup> )	Breastfeeding assessment and determine areas for improvement/intervention Provider or Parent	Has been used in full-term and preterm infants	4 items: Latch-on, Audible swallowing, Nipple comfort, Assistance with positioning	Interrater ICC = 0.78-0.89 (Chapman) 85%-100% interrater agreement (Adams) Test-retest $r = 0.78$ (Riordan)	Construct validity with IBFAT ( $r = 0.69-0.71$ ), and MBA ( $r = 0.68-0.88$ ) (Altuntas 2014 & Riordan 1997) Poor predictive validity for maintaining BF after 6 wk ( $r_s < 0.26$ , Riordan 2001), and for maternal satisfaction and breastfeeding problems ( $r_s = 0.06-0.5$ , Altuntas 2014)
Breastfeeding Self-Efficacy-Short Form (Dennis, 2003 <sup>61</sup> )	Measurement of breastfeeding confidence Breastfeeding women	Healthy, full-term infants	14 items: maternal confidence	Cronbach alpha = 0.89 (Wutke, 2007-0.97, Dennis, 2003 <sup>61</sup> )	Predictive validity, with significant differences in score between breastfeeding and bottle-feeding women ( $P < .001$ , Dennis, 2003 <sup>61</sup> ) Predictive validity, with higher scores postpartum correlated with EBF at 8- and 16-wk postpartum ( $P = .003$ , Wutke, 2007) Construct validity with significant differences in scores between women with and without prior BF experience ( $P < .001$ , Dennis, 2003 <sup>61</sup> ) and women with more children ( $r_s = 0.19$ , $P = .024$ , Oliver-Roig, 2012) Correlated with global self-efficacy index ( $r = 0.41$ , $P < .001$ ) and Stress Management Self-Efficacy Scale ( $r = 0.24$ , $P = .005$ , Oliver-Roig, 2012)
i-GERQ-R (Kleinman 2006 <sup>63</sup> )	Evaluation of GERD symptom severity and treatment effectiveness Parent	Healthy infants	12 items: frequency, amount and discomfort with spit up, refusal/stopping feeding, crying/fussing, hiccups, arching back, stop breathing/colour changes	Cronbach alpha = 0.86-0.87 ICC = 0.85 for test-retest reliability ICC = 0.63 for interrater reliability	Significantly difference scores between control and infants with GERD ( $P < .01$ ) Correlated with caregiver daily symptom diary ( $P < .001$ )



review.<sup>34,35,41</sup> Sample sizes in these three studies were higher, ranging from 54 to 237. BSES-SF was administered pre-frenotomy in all three studies. Emond et al<sup>41</sup> administered the tool again at 5-days and 8-weeks post-procedure and found statistically significant improvements in scores from baseline to day 5 post-procedure ( $P = .002$ ), with no differences between groups at the 8-week measurement, noting that most infants in the control group had undergone frenotomy by this time point. Breastfeeding self-efficacy improved in mothers whose infants underwent frenotomy, with fewer infants being fed by bottle by the 5-day follow-up compared with the control group. The two studies conducted by Ghaheri et al evaluated BSES-SF using the short-form immediately prior to frenotomy and again at 1-week and 1-month post-procedure. Both studies demonstrated statistically significant improvements in scores at 1-week and 1-month time points, compared with pre-procedure scores ( $P < .001$ ).<sup>34,35</sup>

### 3.3 | Infant gastroesophageal reflux

The two studies by Ghaheri et al<sup>34,35</sup> evaluated scores on the Infant Gastroesophageal Reflux Questionnaire—Revised (I-GERQ-R) pre-frenotomy and 1-week and 1-month post-frenotomy. Gastroesophageal reflux disease (GERD) is defined by the movement of gastric contents from the stomach to the oesophagus that causes bothersome symptoms and impairs daily functioning.<sup>49</sup> Both studies that evaluated GERD revealed significant decreases in symptoms of gastroesophageal reflux at all time points after frenotomy ( $P < .001$ ). Factors that may have led to improvement in GERD beyond frenotomy, such as dietary changes in the breastfeeding mother, use of acid-reduction medications or maturation of the infant gastrointestinal tract, were not described.

### 3.4 | Psychometric properties of outcome measures

To address the second aim of this paper, the quality of measures used to assess symptoms of problematic feeding was evaluated.

#### 3.4.1 | Nipple pain

Standardised tools used to assess nipple pain included the VAS,<sup>50</sup> the NRS<sup>51</sup> and the SF-MPQ.<sup>52</sup> These three pain scales have been used in research for both acute and chronic pain, most commonly for the assessment of back pain, but have not been validated for use to measure maternal nipple pain.

#### 3.4.2 | Breastfeeding assessment

Three instruments were used to evaluate breastfeeding, the IBFAT,<sup>48</sup> the LATCH tool<sup>53</sup> and the BBAT.<sup>54</sup> The psychometric properties of these instruments were reviewed in the literature and have been summarised on Table 2.

The IBFAT consists of four items: readiness to feed, rooting, fixing and sucking.<sup>48</sup> It was developed to assess breastfeeding readiness and competence of infants and can be used by mothers and healthcare professionals. Scores range from 0 to 12, with 12 indicating the most effective feeding. Higher IBFAT scores have been associated with greater satisfaction with breastfeeding from the maternal perspective.<sup>55</sup>

The LATCH is a tool used to document breastfeeding assessment, focusing on the key areas of breastfeeding using the LATCH acronym: Latch, Audible swallowing, Type of nipple, Comfort (breast/nipple) and Hold (positioning).<sup>53</sup> Scores range from 0 to 10, with scores of nine or ten representing a breastfeeding session that does not require intervention. The LATCH tool can be completed by mothers or health care providers assessing a breastfeeding session. Both the LATCH tool and the IBFAT have been criticised for their use in evaluating breastfeeding pre- and post-frenotomy, considered to not be specific enough to measure effects of tongue tie or frenotomy on breastfeeding.<sup>56</sup>

The BBAT was developed for use by breastfeeding support providers to evaluate breastfeeding proficiency specifically for mothers with tongue-tied infants.<sup>57</sup> The BBAT is a 4-item tool measuring positioning, attachment to the breast, sucking and swallowing. Scores range from 0 to 8, with higher scores indicating greater proficiency with feeding. At the time the tool was developed, it was found to be correlated with the Breastfeeding Self-Efficacy Scale (BSES) and to have acceptable internal consistency and interrater reliability (Table 2).<sup>54</sup>

#### 3.4.3 | Breastfeeding self-efficacy

Self-efficacy was originally defined by Bandura<sup>58</sup> as “an individual's confidence in his or her perceived ability to perform a specific task or behaviour”. Dennis developed the self-efficacy framework specific to breastfeeding confidence<sup>59</sup> and later developed the BSES<sup>60</sup> and then the short form (BSES-SF).<sup>61</sup> Scores on the BSES-SF range from 0 to 70, with higher scores indicative of greater breastfeeding efficacy and confidence from the maternal perspective.

#### 3.4.4 | Gastroesophageal reflux

The Infant Gastroesophageal Reflux Questionnaire (I-GERQ),<sup>62</sup> and its revised form, the I-GERQ-R<sup>63</sup> measure symptoms of gastroesophageal reflux. The I-GERQ-R is a 12-item questionnaire, with item responses based on a 1-week recall. Higher scores indicate worse symptoms.

## 4 | DISCUSSION

This systematic review identified 20 research studies that compared symptoms of problematic feeding before and after frenotomy in

infants under 1 year old with tongue tie. The literature suggests that there are improvements in maternal symptoms such as nipple pain, LATCH scores and self-efficacy with breastfeeding after frenotomy, with greater improvements when frenotomy is performed before 1 month of age. Frenotomy is a minimally invasive procedure without major complications noted, and maternal improvements following the procedure were present in all of the studies reviewed. However, there are significant limitations to the current literature that restrict our understanding of who benefits most from frenotomy, which methods of treatment should be used, and in what ways the infant may benefit from frenotomy with regards to feeding.

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

The studies included in this review did not consistently and comprehensively describe the study samples in a way to allow readers to understand to whom their results apply. It is unclear what subset of the population seeks treatment for tongue tie, where referrals are generated, and if there are socioeconomic differences within the population of infants with tongue tie. Emond et al noted a difference between the population of mother-baby dyads delivering at the hospital where data were collected and those who presented to the breastfeeding clinic but did not specifically describe these differences. National data in the United States have demonstrated that more diagnoses of tongue tie and treatment with frenotomy occur in families with private insurance and in mid- to high-income areas.<sup>21</sup> Sociodemographic factors of the mothers and infants were not reported in any of the reviewed studies, a major limitation that fails to recognise societal differences that may be present in a country where universal health care and insurance coverage for frenotomy is not guaranteed.

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

infant feeding and are important to know when evaluating to whom frenotomy provides benefit.

While experts in the field of tongue tie recommend frenotomy via laser, research has not been conducted comparing the outcomes of treatment between laser and scissors. Two case reports have demonstrated advantages of using laser for frenotomy, including decreased bleeding, improved visibility and greater ability to manipulate the tissue.<sup>64,65</sup> It is reassuring that no complications were reported in any of the reviewed studies, with the majority using scissors to correct tongue tie. Several studies did not report that exact method used for frenotomy, another limitation of the current research.

The literature provides information about only certain aspects by which infants and mothers benefit from frenotomy with regards to feeding. Knowing that maternal nipple pain is one of the most frequent reasons for breastfeeding cessation, it is clear why this was a commonly reported outcome. Across studies, an improvement in nipple pain was reported. The findings by Khoo et al<sup>37</sup> suggest that resolution of nipple pain, as a direct complication of tongue tie, has the greatest impact on long-term breastfeeding success. However, the reduction of nipple pain cannot be solely attributed to frenotomy, as nipple pain may diminish over time without intervention or may improve with other interventions, such as involvement of a lactation consultant, treatment of thrush or use of a nipple shield. Conversely, nipple pain that results from something unrelated to the infant's tongue tie (eg, untreated thrush) may not improve after frenotomy, even if tongue tie was a contributing factor. Studies exploring the effects of frenotomy on tongue tie need to document assessment of both the infant and maternal anatomy for other contributing factors by trained personnel and report concurrent treatment strategies that may impact the effect of frenotomy on infant feeding.

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

The infant factors that were measured, specifically symptoms of GERD, suck quality and milk transfer, improved. Suck quality and milk transfer were assessed in very small samples and further research on larger samples is needed. Aerophagia-induced gastroesophageal reflux has been associated with poor latching in the presence of tongue tie.<sup>66</sup> These findings should be explored in larger samples, controlling for co-morbidities and variables that may contribute to the development of GERD.

The studies reviewed lack comprehensive evaluation of infant feeding. The infant feeding tools that were used contain very few questions. While this decreases the time it takes to evaluate feeding, they cannot comprehensively assess infant feeding characteristics or difficulties. The LATCH tool has poor predictive validity of breastfeeding problems,<sup>55</sup> differences in scoring between lactation

consultants and mothers,<sup>67</sup> and three of the five assessments are directed at problems experienced by the mother, not the infant (type of nipple, comfort with feeding and hold/infant positioning). The IBFAT has demonstrated poor predictive validity.<sup>48,55</sup> Despite the BBAT being designed to assess problems specifically related to tongue tie, there has not been psychometric testing to fully evaluate this measure. It is difficult to determine when the greatest improvement is seen after frenotomy, with outcome measures being evaluated over varied time spans, both in-person and via telephone follow-up.

#### 4.1 | Recommendations for practice

Despite the limitations of the studies reviewed, breastfeeding assessments and nipple pain improved post-frenotomy. The possibility of tongue tie causing nipple pain and the evidence that frenotomy improves nipple pain should be considered by healthcare providers during feeding assessments and when mothers report prolonged pain with breastfeeding. It is reasonable for healthcare providers to add tongue tie as a differential diagnosis when nipple pain is present and persists despite position changes, latch adjustment and/or lactation support. Providers should be trained in the appropriate method for screening and diagnosing tongue tie, with the understanding that an improved screening measure is necessary. Issues with bottle feeding, such as difficulty latching, clicking, dribbling, excessive gas or prolonged feedings also warrant oral assessment. Tongue tie should be considered as a potential factor in the development of infant GERD. With the known adverse effects of acid-reducing medications,<sup>68</sup> an oral assessment to rule out tongue tie as a potential cause of GERD is reasonable prior to initiation of these medications.

#### 4.2 | Recommendations for research

Future research must control for infant and maternal characteristics that may impact feeding, such as history of breastfeeding experience, infant prematurity, current infant age and the presence of co-morbidities known to affect feeding (eg, congenital heart disease, bronchopulmonary dysplasia). A valid, reliable and standardised assessment tool must be developed to improve effectiveness of screening for the anomaly. Maternal report of nipple pain and breastfeeding difficulties is important; however, it is also necessary to conduct research that focuses on infant-specific feeding outcomes related to tongue tie and frenotomy. This can be accomplished by using comprehensive feeding-related outcome measures that evaluate feeding difficulties. The use of a valid and reliable tool to assess problematic feeding would enhance the rigour of the research beyond measuring maternal symptoms or the subjective reporting of symptoms. In addition, the development of an objective, physiologic evaluation for maternal symptoms and breastfeeding success will provide more information on the benefit of frenotomy

for tongue tie without the maternal bias associated with subjective reporting.

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

## 5 | CONCLUSION

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

#### CONFLICT OF INTEREST

The authors have no commercial associations or sources of support that might pose a conflict of interest. The authors have no other situations that might be perceived as a conflict of interest and there are no copyright constraints.

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