




A cross-sectional study of breastfed infants referred for tongue tie assessment and frenotomy in one Canadian health region

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ABSTRACT

Importance: Tongue tie (TT) is a condition that can cause infant feeding difficulties due to restricted tongue movement. When TT presents as a significant barrier to breastfeeding, a frenotomy may be recommended. Universally accepted diagnostic criteria for TT are lacking and wide prevalence estimates are reported. New referral processes and a Frenotomy Assessment Tool were implemented in one Canadian health region to connect breastfeeding dyads with a provider for TT evaluation and frenotomy.

Objective: To determine the proportion of babies with TT as well as the frequency of frenotomy.

Methods: This cross-sectional study included infants who initiated breastfeeding at birth and were referred for TT evaluation over a 14-month period. Data were collected retrospectively by chart review and analyzed using SPSS. Factors associated with frenotomy were examined using logistic regression.

Results: Two hundred and forty-one babies were referred. Ninety-two percent ($n = 222$) were diagnosed with TT and 66.0% ($n = 159$) underwent frenotomy. In the multivariate model, nipple pain/trauma, inability to latch, inability to elevate tongue, and dimpling of tongue on extension were associated with frenotomy ($P < 0.05$). Most referrals in our region resulted in a diagnosis of TT; however, the number of referrals was lower than expected, and of these two-thirds underwent frenotomy.

Interpretation: TT is a relatively common finding among breastfed infants. Future research should examine whether a simplified assessment tool containing the four items associated with frenotomy in our multivariate model can identify breastfed infants with TT who require frenotomy.

KEYWORDS

Breastfeeding, Nutrition, Community health services, Pediatrics

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INTRODUCTION

Ankyloglossia, commonly known as tongue tie (TT), is an anatomic variation where the membrane under the tongue (i.e., the lingual frenulum) is short, inelastic, thickened, or attached too close to the tongue tip.^{1,2} These anatomical findings can be identified on inspection and palpation and may impair tongue function during breastfeeding.^{3,4} An infant with TT may have trouble attaching to the breast and/or maintaining an effective latch and suckle; TT can lead to complex breastfeeding difficulties and in some cases early cessation of breastfeeding.^{2,5} As such, TT poses an important threat to the longevity of breastfeeding exclusivity and its associated health benefits. Exclusive breastfeeding for the first six months of life is recommended by the Canadian Pediatric Society and the World Health Organization (WHO) due to its protective health effects for both the infant and mother.^{6–8} From a population health perspective, it is important to establish comprehensive healthcare services that include support for breastfeeding dyads who may be experiencing feeding difficulties due to TT.

There are no universally accepted criteria for the diagnosis of TT, and there is continued debate in the medical community about the appropriateness of frenotomy—a procedure whereby the lingual frenulum is cut or divided.⁹ The use of inconsistent diagnostic criteria and continued debate on this topic has likely contributed to the wide variation in the reported prevalence of TT where population-based estimates range from 4% to 11%.^{9,10} Furthermore, the overall prevalence of TT is directly influenced by the tool used for diagnosis.¹¹ Increases in support for breastfeeding initiation and the Baby Friendly Hospital Initiative may have contributed in a substantial way to the increased rates of TT diagnosis in Canada and beyond.¹² Similar trends have been noted for treatment, where “runaway rates for frenotomy” have been described in some parts of Canada.¹³

Despite the lack of agreement on criteria for diagnosing TT, several tools have been developed by clinician-scientists throughout the world to aid the assessment of TT and its severity, and/or to determine the need for frenotomy. One such tool that has undergone validity and reliability testing is the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF);^{14,15} however, this tool is not without limitations. It has been argued that the length and complexity of the HATLFF make it challenging to use in clinical practice.^{16,17} A newer tool, the Frenotomy Decision Tool for Breastfeeding Dyads, contains fewer items and involves an evaluation of both mother and baby during a breastfeeding session.^{2,18} The choice of tool is an important one since it can have a direct impact on the identified prevalence of TT in a population.¹¹

We became interested in studying the epidemiology of TT in breastfeeding infants because of clinical experience, low provincial exclusivity rates at six months, and a desire to support breastfeeding families in our Canadian province. Breastfeeding initiation rates increased in our province from 64% in 2007 to 76% in 2017; however, exclusive breastfeeding to 6 months remains low at 15%.¹⁹ To this end, our research team is committed to studying breastfeeding practices, attitudes, and behaviors in the province.^{5,20–25} Moreover, the results of our qualitative study on mothers' experiences breastfeeding a child with TT highlighted the inadequate referral, assessment, and treatment processes for TT in our health region.⁵

Recognizing the need for an evidence-informed approach to assist families experiencing breastfeeding challenges, in 2016 we held a nationally-funded research planning and knowledge exchange forum on the topic of ankyloglossia and infant nutrition. This meeting brought together clinicians, researchers, policymakers, and citizens with lived experience. Through discussion, the need for a publicly funded, systematic, and coordinated approach to TT assessment, diagnosis, and management from birth and throughout infancy was identified. One major output of the meeting was the development of a Frenotomy Assessment Tool and a referral pathway in the community (i.e., outpatient) setting. This work was led by a clinician-scientist (Jessica Bishop) on our research team and implemented within the eastern region of our province, serving approximately 210 000 people in the census metropolitan area.²⁶

In accordance with institutional policies and procedures, the Frenotomy Assessment Tool was constructed by a team of experienced health professionals and reviewed by the regional forms committee. The tool used in our region was informed by the Frenotomy Decision Tool for Breastfeeding Dyads and is intended to help identify those babies who may benefit from a referral to a provider who is trained to perform an in-depth assessment of TT.^{2,18} The tool consists of two components: (1) a functional assessment of the infant feeding at the breast; and (2) visual inspection and/or palpation of the lingual frenulum and tongue. Where an infant scores one or greater on the functional assessment and two or greater on the visual assessment, a referral is initiated. Prior to enacting the new referral pathway on July 1, 2017, the second author (Jessica Bishop) trained public health nurses and lactation consultants working in the community on the appropriate use of the assessment tool; case examples, and a demonstration of the functional and visual assessment processes were central to this training. A recording of the training session was also made available for future reference.

Newborns in our hospital who are experiencing significant breastfeeding challenges within the first days of life and who have a visible or palpable TT are offered a referral for frenotomy. These referrals are initiated by the attending physician (i.e., pediatrician or family physician) or lactation consultant after a breastfeeding assessment is performed. Referrals in the hospital are sent to the pediatric dentist, or on rare occasions, an otolaryngologist. Prior to 2017, TT assessment was not a regular component of newborn screening in our facility. However, through knowledge exchange and a review of best practices, opportunities to re-examine newborn screening were created. Today, a more comprehensive assessment of infant feeding at the breast is undertaken prior to discharge and the orofacial structures are examined thoroughly.

To ensure wrap-around services are available, in the community or outpatient setting, TT referrals are sent to one of three public providers (pediatric dentist, family physician, or an otolaryngologist). Consideration is given to the age of the infant and clinical complexity when the referral is initiated. Infants less than 12 weeks old are referred to the family physician, those greater than 12 weeks old are referred to the pediatric dentist, and complex presentations are referred to the otolaryngologist. Public health nurses and lactation consultants working in publicly funded community breastfeeding clinics are the primary sources of support for breastfeeding dyads and would most commonly initiate referrals for TT assessment and management using the Frenotomy Assessment Tool described previously. All providers included within the inpatient (i.e., hospital) and outpatient (i.e., community) referral pathways have experience performing frenotomies and using scissors to cut the lingual frenulum.

Implementation of the Frenotomy Assessment Tool and TT referral process within our health region was initiated for the primary purpose of supporting dyads who are experiencing breastfeeding difficulties due to TT. It was our hope that this new care process would improve breastfeeding outcomes while simultaneously decreasing unnecessary interventions for TT, specialist consults, and health care utilization costs. From a research perspective, the implementation of the new referral process allowed us to gain a better understanding of the epidemiology of TT and its impact on breastfeeding outcomes in our region. This work presents the findings of our research following the first 14 months of implementation. The primary objective of our study was to describe the proportion of breastfed babies presenting with TT who required frenotomy and factors statistically associated with frenotomy in our sample.

METHODS

Ethical approval

Ethics approval for this study was obtained from the Newfoundland and Labrador Health Research Ethics Board, Reference No. 18.055.

Study design and sample

We conducted a cross-sectional study of infants born at our provincial pediatric hospital who initiated breastfeeding and were referred to an affiliated provider for TT assessment and frenotomy between July 1, 2017, and August 31, 2018. All infants referred via the inpatient (i.e., in the first days of life, before discharge) or outpatient (i.e., community) route were included in this study. The sample size calculated for this study was 220, using the formula for determining the frequency of a factor in the population with a population size of approximately 2000 babies born annually who initiate breastfeeding, 20% as the anticipated frequency of TT, a 5% margin of error, and 95% confidence level.²⁷ We decided to use 20% as the anticipated frequency of TT in calculating sample size for three reasons: the observed prevalence of TT is directly influenced by the assessment tool used to make a diagnosis, where values range from 2% to 20%¹¹; a rapid temporal increase in newborn TT and frenotomy was observed in our province from 2010 to 2014¹²; and we had observed a growing public awareness of TT and frenotomy within local breastfeeding support groups and social media sites.

Data collection

Data were abstracted retrospectively by an experienced research nurse/research assistant through chart review. Data from the medical records of both mother and baby were retrieved using data collection forms that had been developed by the research team a priori and approved by the local health ethics board. Demographic variables for the infant (e.g., sex, gestational age, and birth weight) and mother (e.g., age, gravida, and previous breastfeeding experience) were collected as well as birth procedures, route of TT referral (i.e., inpatient versus outpatient), infant feeding practices and breastfeeding challenges at the time of referral, visual and functional characteristics of the infant's tongue and feeding, the type of frenotomy procedure performed and its complications (where applicable), and clinical outcomes at 2 weeks following frenotomy (e.g., improvement in symptoms and feeding practices). To ensure the accuracy of data collection, 10% of the charts were reviewed by the principal investigator (first author, Tiffany A. Lee).

Newborns referred to the pediatric dentist or an otolaryngologist via the inpatient route were identified by two mechanisms: (1) upon review of the referral logbooks maintained by nursing staff on the obstetrics unit and (2) through electronic patient records maintained by dentistry; duplicates were merged accordingly. We were not able to perform an additional search of electronic patient records maintained by the otolaryngologist's office, as there is no specific diagnostic or billing code for TT or frenotomy in that system. That said, a small number of otolaryngologist referrals were identified using the obstetrical logbooks. Furthermore, staff at the otolaryngologist's office indicated that only a small number of babies were seen in their office. Data sources for the inpatient referrals included 1) dentistry electronic patient records; and 2) electronic medical records (EMRs) maintained by the health authority.

Infants referred via the outpatient route (i.e., the community referral pathway) were identified by diagnostic codes within each clinic's electronic health record. Data sources for community referrals included 1) electronic patient charts maintained by the provider; and 2) EMRs maintained by the regional health authority. A list of community referrals to the otolaryngologist's office could not be retrieved for the same reasons noted previously.

Data analysis

Data were analyzed using the Statistical Package for the Social Sciences (Version 28). Descriptive statistics included calculation of mean and standard deviation (SD) for continuous variables (age of infant and mother, gestational age, infant weight at birth, etc.) and frequencies and percentages for categorical variables (sex of infant, gravidity, etc.). First, univariate logistic regression techniques were used to examine the relationship between the functional and visual characteristics of infant feeding (i.e., independent variables) and frenotomy (i.e., binary dependent variable). Significant factors were identified at a level of $P < 0.05$ in the univariate analysis. Next, multivariate logistic regression with a backward stepwise selection method was used to determine the final model and adjusted for the following variables: infant sex, gestational age, and birth weight. Odds ratios with associated 95% confidence intervals are reported, and statistical significance was $P < 0.05$. The final model classification index (C-Index) was also reported.

RESULTS

Demographics

The number of live births at our provincial pediatric hospital from July 1, 2017, to August 31, 2018, was 2647, and of these, 76.2% of infants initiated breastfeeding. The total number of infants included in this study who

TABLE 1 Characteristics of study population ($n = 241$)

Variables	Number
Maternal characteristics	
Age (year)	31.8 \pm 4.7
Multigravida	143 (59.3)
Previous breastfeeding experience [†]	103 (42.7)
Previous child with tongue tie [‡]	32 (13.3)
Flat or inverted nipples	26 (10.8)
Delivery characteristics	
Cesarean	72 (29.9)
Vaginal	169 (70.1)
Infant characteristics	
Male	122 (50.6)
Gestational age (week)	39.2 \pm 1.4
Birth weight (g)	3444.9 \pm 504.3
Hypoglycemia after delivery [¶]	19 (7.9)
NICU admission after delivery	20 (8.3)
Feeding characteristics [§]	
Exclusive breastfeeding	100 (41.5)
Breastfeeding plus supplementation	110 (45.6)
Bottle feeding only (expressed breast milk)	27 (11.2)

Data are shown as n (%) or mean \pm SD.

[†]Missing data = 3; [‡]Missing data = 21; [§]Missing data = 3;

[¶]Hypoglycemia, blood glucose < 2.6 mmol/L

were referred for evaluation of TT and frenotomy was 241. The mean gestational age of infants was 39.2 \pm 1.4 weeks and 50.6% ($n = 122$) of babies were assigned male sex at birth. The average maternal age was 31.8 \pm 4.7 years and 42.7% ($n = 103$) of mothers had prior experience breastfeeding an infant. Further details regarding the characteristics of the study population are presented in Table 1.

Referrals, diagnosis, and frenotomy

Of the 241 babies included in this study, 36.9% ($n = 89$) were referred before hospital discharge (i.e., inpatient route) and 63.1% ($n = 152$) were referred by the outpatient route (i.e., community-based referrals). Of the inpatient referrals, 57.3% ($n = 51$) were initiated by pediatricians, 39.3% ($n = 35$) by lactation consultants, and 3.4% ($n = 3$) by family physicians. In contrast, 63.8% ($n = 97$) of outpatient referrals were initiated by public health nurses, 25.7% ($n = 39$) by family physicians, 9.9% ($n = 15$) by lactation consultants, and 0.7% ($n = 1$) by pediatrician. Two hundred and twenty-two infants, or 92.1%, were diagnosed with TT. From that number, 49.5% ($n = 110$) of babies had an anterior tie and 50.5% ($n = 112$) had a posterior tie. These findings suggest that the prevalence of

TABLE 2 Functional and visual characteristics of infant feeding and their relationship with frenotomy

Variables	n (%)	Odds ratio (95% CI) [†]	P [†]	Odds ratio (95% CI) [‡]	P [‡]
Functional assessment					
Nipple pain or trauma	164 (68.0)	2.657 (1.511–4.674)	<0.001	2.009 (1.055–3.824)	0.034
Inability to latch	178 (73.9)	2.896 (1.599–5.244)	<0.001	2.649 (1.330–5.276)	0.006
Poor milk transfer	87 (36.1)	1.457 (0.826–2.572)	0.194	-	-
Poor weight gain	53 (22)	1.004 (0.527–1.910)	0.991	-	-
Clicking sound while sucking	30 (12.4)	0.876 (0.395–1.941)	0.744	-	-
Visual assessment					
Inability to elevate tongue midway to the roof of the mouth	115 (47.7)	3.234 (1.829–5.719)	<0.001	2.177 (1.131–4.190)	0.020
Inability to protrude tongue past gum line	98 (40.7)	3.601 (1.960–6.16)	<0.001	-	-
Inability to maintain suction at the breast	93 (38.6)	3.175 (1.727–5.834)	<0.001	1.969 (0.993–3.905)	0.052
Dimpling of the tongue on extension	51 (21.2)	6.269 (2.393–16.493)	<0.001	5.048 (1.801–14.149)	0.002

[†]Univariate analysis; [‡]Multivariate analysis, and adjusted for the following variables infant sex, gestational age, and birth weight. Abbreviations: CI, confidence interval; -, not applicable.

TT among breastfed babies in our region is approximately 11% (i.e., 222 of 2016 babies who initiated breastfeeding at birth).

Two-thirds, or 66.0% ($n = 159$), of babies who were referred underwent frenotomy. The mean age of infants referred by the outpatient route, at the time of frenotomy was 42.2 ± 28.7 days. Eighty-four of the 159 (52.8%) infants who underwent frenotomy attended a follow-up appointment at 1–2 weeks post-frenotomy. The majority, 83.3% ($n = 70$) of parents reported improvements in symptoms at follow-up and 97.6% ($n = 82$) of infants experienced no complications after frenotomy. One baby had a second frenotomy procedure and another baby had an emergency room visit.

Factors associated with frenotomy

As described previously, the assessment process involved an examination of infant feeding at the breast by one or more trained healthcare professionals. Feeding difficulties documented in the infant's chart by the healthcare professional(s) who completed the assessment are provided in Table 2. In terms of the functional assessment, nipple pain/trauma and inability to latch were statistically associated with frenotomy in the univariate analyses. There was also a statistical association between frenotomy and each of the four visual characteristics (Table 2). Nipple pain/trauma, inability to latch, inability to elevate tongue, and dimpling of the tongue on extension achieved statistical significance in the final multivariate model (Table 2); the backward logistic selection method removed the other four variables in the SPSS analysis either due to multicollinearity or they did not achieve statistical significance in the final model. The final model C-Index was 78.8%.

DISCUSSION

The findings of our study suggest that TT is a relatively common finding among breastfed babies with an estimated prevalence of 11% in our sample. As such, the prevalence of TT in our region sits at the higher end of population-based estimates reported by organizations like the Canadian Paediatric Society and the American Academy of Pediatrics.^{10,28} However, it should be noted that these estimates are based on the results of studies published in the early 2000s and do not necessarily reflect current healthcare practice; more recent estimates of TT in infants range from 2% to 20% depending on the assessment tool used.¹¹

The importance of providing multidisciplinary, wrap-around services for mother-infant dyads who are experiencing breastfeeding difficulties due to TT cannot be overstated. The majority of infants referred for evaluation in our study were diagnosed with TT; however, the number of referrals was lower than expected, and not all babies required frenotomy. These findings highlight the critical need for a coordinated and timely approach to breastfeeding support, beginning in the hospital, and continuing in the community. Furthermore, with appropriate levels of breastfeeding support, it has been estimated that anywhere from 40%–75% of babies with TT will be able to breastfeed successfully.⁴ In our health region, the implementation of a new referral pathway and the Frenotomy Assessment Tool, plus educational programming for public health nurses, as well as a small number of frenotomy providers, may have improved the consistency of care and contributed to lower numbers of referrals and interventions than expected. Dixon and colleagues took a similar approach in New Zealand with the

development and implementation of a multifaceted program for the assessment and management of TT among dyads who were experiencing breastfeeding difficulties.²⁹ They reported substantial reductions in the rate of frenotomy following the implementation of their clinical pathway, as well as successful uptake of the program by health professionals.²⁹

There are several TT assessment tools that can be used in conjunction with a structured breastfeeding assessment to determine the need for frenotomy, though none are universally accepted in practice. The HATLFF has been recommended by the Academy of Breastfeeding Medicine;^{15,30,31} however, the length and complexity of the tool make it challenging to apply across all practice environments.^{16,17} In our health region, a decision was made to develop a new tool—the Frenotomy Assessment Tool, which was informed by the Frenotomy Decision Tool for Breastfeeding Dyads.^{2,18} The results of our study suggest that the tool does a good job of identifying babies with TT who may benefit from frenotomy. As a result, the use of the tool has been extended to other areas of the province and two additional family physicians have joined the team of frenotomy providers. However, it may be possible to reduce the number of items included in our Frenotomy Assessment Tool, as only four factors (nipple pain/trauma, inability to latch, inability to elevate tongue midway to the roof of the mouth, and dimpling of tongue on extension) were statistically associated with frenotomy in the multivariate model. A shortened tool may in fact improve diagnostic consistency and implementation across busy breastfeeding clinics as well as reduce unnecessary referrals for frenotomy and surgical interventions. That said, prior to large-scale implementation of such a tool, validity and reliability testing must be performed. In addition, longitudinal studies are needed to better understand whether frenotomy improves breastfeeding outcomes and exclusivity at six months.

This study has strengths and limitations. To our knowledge, this is the first observational study examining the frequency of TT and frenotomy following the implementation of multidisciplinary wrap-around services for breastfeeding dyads in Canada. A strength of our study relates to the inclusion of all infants referred for evaluation of TT (i.e., newborns referred before hospital discharge and infants referred by providers in the community) in the study sample, providing a more complete picture of TT and frenotomy over the infancy period. Although we did not perform inter-rater reliability testing on the use of the Frenotomy Assessment Tool by public health nurses at the time of referral, the assessment, diagnosis, and management of TT were carried out by a very small number of providers, improving diagnostic consistency and reliability of findings. Our inability to include referrals to ENT is a limitation. It

is also possible that additional breastfeeding dyads may have sought out frenotomy from providers working in private practice and therefore were not included in our study; however, the total number is expected to be quite low since there would be out-of-pocket costs associated with obtaining care from private practice. Finally, it should be acknowledged that our sample is representative of babies who were well, born at full-term to mothers older in age, and breastfeeding. As such, it could be argued that our sample is more representative of a higher socioeconomic status population and may not reflect true population estimates of TT and frenotomy among infants.^{32,33}

To conclude, the findings of our research suggest the prevalence of TT among breastfed babies in our region is approximately 11%. The majority of infants referred for evaluation were diagnosed with TT and frenotomy was performed in two-thirds of cases. Wrap-around services—beginning in the hospital and continuing in the community—for mother-infant dyads experiencing breastfeeding difficulties due to TT, can help to identify infants who may benefit from frenotomy. A comprehensive symptom assessment should be paired with the functional and visual assessment of the infant feeding at the breast, as TT may not always be obvious on inspection. Future research should examine whether a simplified assessment tool, containing the four items associated with frenotomy in our multivariate model, can be validated in other populations and settings and used to identify breastfed infants with TT who require frenotomy. A shortened frenotomy assessment tool may lead to better diagnostic standards and uptake by clinicians.

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Please contact the corresponding author should you be interested in obtaining a copy of the Frenotomy Assessment Tool used within our institution.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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