Nipple Pain at Presentation Predicts Success of Tongue-Tie Division for Breastfeeding Problems

Authors A. K. K. Khoo, N. Dabbas, N. Sudhakaran, N. Ade-Ajayi, S. Patel

Affiliation King's College Hospital, Department of Paediatric Surgery, London, United Kingdom

Key words
- tongue-tie
- ankyloglossia
- breastfeeding
- nipple pain

Abstract

Introduction: In mother-infant pairs experiencing breastfeeding difficulties, frenulotomy for tongue-tie may improve breastfeeding. We tested the hypothesis that those experiencing nipple pain are most likely to benefit from the procedure in a prospective cohort study.

Materials and Methods: Mother-infant pairs attending a dedicated clinic for the assessment and treatment of tongue-tie completed a standardised, structured symptom questionnaire. Three months later outcome was assessed by questionnaire. Multivariate logistic regression analysis was used to determine preoperative predictors of successful outcome.

Results: Sixty-two infants <90 days old underwent frenulotomy and completed follow-up. At presentation, 52 mothers (84%) reported nipple pain, and 32 mothers (52%) nipple trauma. Three months after frenulotomy, 78% of respondents were still breastfeeding. Feed lengths (mean reduction: 17 mins; $p<0.001$) and time between feeds (mean increase: 38 mins; $p<0.001$) had significantly improved, as had difficulty of feeding (mean improvement in self-rated difficulty score: 42%; $p<0.001$). Those having difficulty breastfeeding due to nipple pain showed a significant long-term benefit from frenulotomy; pre-frenulotomy nipple pain was associated with an increased likelihood of breastfeeding at 3 months in adjusted multivariate analysis (OR 5.8 [95% CI 1.1–31.6]).

Conclusion: Mother-infant pairs with tongue-tie and breastfeeding difficulties due to nipple pain are most likely to benefit from frenulotomy.

Introduction

Tongue-tie (ankyloglossia) is a congenital condition characterised by a short lingual frenulum which results in tethering of the tongue. The prevalence of tongue-tie in newborn infants varies between 2.5% [1] and 11% [2], depending on the population and the diagnostic criteria used. The majority of infants with tongue-tie will breastfeed without difficulty, but a reported 25% [3] to 44% [4] of these babies have both tongue-tie and breastfeeding difficulties (TBD). Although the evidence is limited, the few studies which have evaluated the effectiveness of frenulotomy for TBD [5,6] have shown it to be a safe procedure, which can improve breastfeeding. In 2005, a specialist committee conducted a review of frenulotomy for TBD on behalf of the National Institute for Health and Clinical Excellence (NICE), in order to determine the role of the procedure in the NHS. The interventional procedure guidelines subsequently issued concluded that the "evidence is adequate to support the use of the procedure provided that normal arrangements are in place for consent, audit and clinical governance" [7].

Although frenulotomy has a risk of only minor complications [7], it is nevertheless important to try and identify which mother-infant pairs are likely to benefit from the procedure in order to ensure that parents are appropriately counselled, that parental expectations of the procedure are realistic and that finite NHS resources are deployed according to a utilitarian principle. As yet, no predictors of success of frenulotomy have been identified in infants with TBD, and indications for frenulotomy remain a matter of debate.

The severity of breastfeeding difficulty does not correlate with anatomical severity of tongue-tie [6]. The Hazelbaker Assessment Tool for Lingual Frenulum Function is a functional score, which identifies babies at risk of breastfeeding difficul-

received May 25, 2009
accepted after revision June 30, 2009

Bibliography
DOI 10.1055/s-0029-1234041
Published ahead of print: 11 September 2009
Eur J Pediatr Surg 2009; 19: 370–373 © Georg Thieme Verlag KG Stuttgart · New York
ISSN 0939-7248

Correspondence
Shailesh Patel
Deparment of Paediatric Surgery, King's College Hospital
Denmark Hill, London, UK
SE5 9RS
shailesh.patel@kch.nhs.uk

ties by physical examination. In infants classified as "function impaired", the author recommends undertaking early tongue-tie division [8]. However, whether Hazelbaker "function impaired" infants derive a benefit from frenulotomy remains unreported.

Maternal nipple pain has been well described as a deterrent to breastfeeding [4,5,9]. Whilst nipple pain occurs in an estimated 60–80% of all breastfeeding mothers during the early postpartum period, in the majority of normal infants this is a transient symptom which peaks on day 3, and resolves within 2 weeks. However, in infants with tongue-tie, maternal nipple pain persists beyond this period in 36–80% of cases [10], and occurs significantly more frequently than in normal infants [3,11]. Frenulotomy for TBD can result in decreased nipple pain [6,12]. In a single blinded study of 25 mother-infant pairs, Dollberg et al. demonstrated an immediate reduction in nipple pain during feeding after frenulotomy compared to a sham procedure [5]. It was not reported whether this resulted in continuation of breastfeeding.

We therefore investigated whether the presence of nipple pain at presentation would identify mother-infant pairs most likely to benefit from frenulotomy, as measured by persistence of breastfeeding at 3 months, with a prospective cohort study.

Materials and Methods

Study Subjects and Design

From June 2007 to July 2008, all mothers of babies <90 days old attending the TBD clinic at King's College Hospital were invited to complete a standardised structured symptom questionnaire. Mother-infant pairs were reviewed in this clinic only after an appropriate period of intervention from qualified lactation consultants had failed to improve feeding difficulties. The questionnaire gathered data on infant characteristics (gender, birth weight, age at attendance and whether there was a family history of tongue-tie), feeding pattern (length of feeds, number of feeds a day, length of time between feeds) and infant and maternal symptoms (presence of nipple pain, nipple trauma, poor latch, noisy feeding, infant frustration and prolonged jaundice). Mothers were asked to rate how difficult they found feeding overall on a Likert scale ranging from 0 to 10, with 0 being "no difficulty feeding" and 10 being "extremely difficult". All infants were examined for tongue-tie by a consultant paediatric surgeon (SP), and the length of lingual tethering gauged by eye and described (extending for 25%, 50%, 75% or to the tip of the full length of the tongue). Whether the baby was able to protrude his tongue beyond the lower gums, or elevate it was noted as basic markers of function.

When symptomatic tongue-tie was confirmed on examination, division of tongue-tie was offered. Following appropriate parental counselling, informed consent was obtained and frenulotomy undertaken in the clinic during the same visit. Mothers had been instructed to withhold feeds for at least one hour prior to clinic. The baby was swaddled securely in a blanket, and held in position by an assistant. The baby's head was fixed firmly by the assistant's left hand, and the baby's chin depressed with the thumb of the right hand to open the mouth. The surgeon then undertook frenulotomy: the tongue-tie was put on a stretch using a sterile notched tongue elevator, and divided using curved blunt strabismus scissors. Completeness of division was confirmed by blunt finger-tip dissection to ensure that a diamond-shaped defect was produced. No anaesthesia or analgesia was required.

Immediately after the procedure, the baby was unwrapped, soothed and taken to the waiting mother to be put immediately to the breast. This comforted the baby, and, in the majority of cases, compression of the tongue against the breast was enough to stop any minor bleeding. Breastfeeding counsellors supported the mother during this feed, offering help and advice. The baby completed the feed, and following re-examination to ensure haemostasis had been achieved, discharged home. Mothers were advised to attend for further breastfeeding support with their referring lactation consultant within the next few days. The mother was asked to complete a follow-up questionnaire detailing feeding patterns and maternal assessment of breastfeeding difficulty at three months, and return it to us by post. Data collection was approved as part of an ongoing clinical service evaluation.

Analysis and Statistics

Changes in feeding patterns were analysed using paired t-tests. Success of the frenulotomy was defined as the mother-infant pair breastfeeding at 3 months. Variables assessed as potential prognostic markers for success included infant demographics (gender, age, family history, birth weight), anatomical severity of tongue-tie (length of tongue-tie), functional severity of tongue-tie (ability to protrude and ability to elevate) feeding patterns (length of feeds, time between feeds, number of feeds a day), presence of feeding symptoms (nipple pain, nipple trauma, poor latch, infant frustration, noisy feeding and prolonged jaundice). Significantly associated factors in univariate analysis were subject to multivariate logistic regression, with backward stepwise elimination used to construct the final model. Values which were normally distributed are given as mean (standard deviation) and p values of <0.05 were considered statistically significant.

Results

Over the 13 month period, 62 mother-infant pairs underwent frenulotomy and completed the 3 month follow-up. Infant characteristics are summarised in **Table 1**. At presentation, 52 (84%) mothers reported nipple pain, and 32 (52%) mothers nipple trauma (see **Table 2**). Feeding patterns in this population were markedly abnormal (see **Table 3**), with feeds lasting for a mean length of 40 min, with some individuals reporting feeds lasting on average 2 hrs. The mean time between feeds was two and a half hours. Mothers found feeding difficult, with a mean rating of 6.1 out of 10 for overall feeding difficulty.

The procedure had a high rate of long-term success, with 78% of mother-infant pairs breastfeeding at 3 months. Individual feeding characteristics improved, with a mean reduction in length of feeds of 17 min and time between feeds increased by 38 min, both of which were highly significant ($p<0.001$). The individual self-rated breastfeeding difficulty scores decreased by 42% following frenulotomy ($p<0.001$), and in 52% of mother-infant pairs breastfeeding problems resolved completely, with mothers reporting a score of 0 ("no difficulty feeding") at follow-up.

Multivariate logistic regression analysis identified two independent factors with a predictive value for the long-term success of frenulotomy for TBD. Mother-infant pairs presenting with nipple pain were significantly more likely to be breastfeeding at

Table 1 Infant characterstics.

Infant Characteristics	Number of infants (n = 62)	(%)
male	42	68
family history of tongue-tie	19	33
tongue movement:		
– able to protrude tongue beyond lower gum line	18	31
– able to elevate tongue	15	24
length of lingual tethering:		
– 25% of tongue	7	22
– 50% of tongue	19	30
– 75% of tongue	15	24
– to tip of tongue	21	34
birth weight/kg (mean ± standard deviation)	3.38 (± 0.45)	
age at attendance/days (mean ± standard deviation)	23.5 (± 17.1)	

Table 2 Symptoms at Presentation.

Symptoms at presentation	Number reporting n = 62 (%)
nipple pain	52 (84)
nipple trauma	32 (52)
poor latch	52 (84)
prolonged jaundice	6 (11)
noisy feeding	39 (63)
infant frustration	50 (81)
infant not satisfied after feed	27 (44)

Table 3 Feeding characteristics.

Feeding characteristics	At presentation	Post-frenulotomy	Significance level (paired t-test)
number of feeds/day	7.4 ± 2.4	6.4 ± 1.5	$p < 0.005$
length of feed/min	41.6 ± 27.5	24.1 ± 17.4	$p < 0.001$
time between feeds/min	161.1 ± 44.3	197.0 ± 45.3	$p < 0.001$
overall difficulty (self-rated score: 0 = no difficulty, 10 = maximum difficulty)	6.1 ± 2.7	1.9 ± 2.6	$p < 0.001$

3 months, OR 5.8 [95% CI 1.1–31.6]. Presence of a family history of tongue-tie, however, decreased the likelihood that a baby was breastfeeding at 3 months OR 0.24 [95% CI 0.06–0.94].

No correlation was seen between the severity of the tongue-tie (anatomical or functional extent) and either maternal perception of breastfeeding difficulty at presentation or the likelihood of breastfeeding at 3 months.

Discussion

It is optimal for babies to be exclusively breastfed for the first 6 months of life [13], and this is reflected in current WHO recommendations [14]. However, the most recent Infant Feeding Survey, conducted by the Scientific Advisory Committee on Nutrition, found that only a quarter of UK infants are breastfeeding at 6 months, with just 1% remaining exclusively breastfed at this time [15]. The Baby Friendly Breastfeeding Hospital initiative, an international strategy coordinated by the WHO and UNICEF, underlines the central role of healthcare services in supporting and promoting breastfeeding [16]. Mother-infant pairs with TBD are in a minority in the general population, but they are a group who can potentially benefit from a specifically targeted intervention. Whilst our study population represents a highly motivated group of mothers, three months after frenulotomy 78% of mother-infant pairs were breastfeeding despite initially struggling. Nationally, only one third of infants are breastfeeding at four months of age [15].

We found that nipple pain was a common breastfeeding problem at presentation. The observed prevalence of 84% is similar to that found in other studies. Hogan et al. report that 80% of their study subjects were experiencing nipple pain at presentation [6], with Dollberg et al. finding it a universal problem amongst their subjects [5]. In both studies, reported rates of nipple pain mirrored rates of poor infant latch, and 45 mothers in our study reported both symptoms. In normal breastfeeding, the nipple, areola and underlying breast tissue are drawn into the infant's mouth, and a seal created between the infant's lips and the breast. Suckling creates an intra-oral vacuum, drawing these tissues into a teat, with the tongue forming a central trough around them [17]. Geddes et al. used ultrasound to study babies with tongue-tie, and noted that instead of latching to all tissues, these infants tend to latch onto the nipple, compressing the tip or base with their tongues whilst breastfeeding. Post-frenulotomy, nipple compression ceased, which corresponded to a reduction in nipple pain, improved latch, and better milk transfer [9]. Their findings show that the symptom of nipple pain is a sign of underlying poor latch. The better latch and milk transfer associated with tongue-tie division would explain why we found that the strongest predictor of long-term success of frenulotomy TBD was nipple pain, with mother-infant pairs reporting pain at presentation being 5.8 times more likely to be breastfeeding at 3 months than those without pain.

At 2.1:1, the ratio of male to female babies with tongue-tie we observed is similar to previous findings [11,12]. Also similar is the observation that tongue-tie is often familial: one third of our study population had a family history of tongue-tie. Our clinic has treated sets of siblings with similar symptoms occurring years apart. Why a family history of tongue-tie should be a negative predictor of success of frenulotomy for TBD, however, is unclear from current published literature. It may be that previous experience with TBD had an impact on the levels of motivation with subsequent offspring in this family.

This study has clear limitations. The study population is highly self-selected: not only was it a highly motivated group of mothers who sought referral to the TBD clinic, but we also relied on the voluntarily completion and return of follow-up questionnaires, which introduces further significant selection bias. Our follow-up questionnaire gathered data regarding feeding methods and patterns at three months, but mothers were not asked about specific symptoms, so we are unable to comment directly on the effect frenulotomy had on these.

Conclusion

Previous trials have established that frenulotomy can be effective for TBD. Our study has shown that mother-infant pairs where the mothers are experiencing nipple pain are particularly likely to benefit from this procedure.

Conflict of interest: None

References

1 *Flinck A*, *Paludan A*, *Matsson L et al*. Oral findings in a group of newborn Swedish children. Int J Paediatr Dent 1994; 4: 67–73
2 *Freudenberger S*, *Santos Diaz M*, *Bravo J et al*. Intraoral findings and other developmental conditions in Mexican neonates. J Dent Child (Chic) 2008; 75: 280–286
3 *Messner A*, *Lalkea M*. Ankyloglossia: incidence and associated feeding difficulties. Arch Otolaryngol Head Neck Surg 2002; 126; 36–39
4 *Griffith D*. Do tongue-ties affect breastfeeding? J Hum Lact 2004; 20: 409–414
5 *Dollberg S*, *Botzer E*, *Grunis E et al*. Immediate nipple pain relief after frenotomy in breast-fed infants with ankyloglossia: a randomized prospective study. J Pediatr Surg 2006; 41: 1598–1600
6 *Hogan M*, *Westcott C*, *Griffiths M*. Randomized, controlled trial of division of tongue-tie in infants with feeding problems. J Paediatr Child Health 2005; 41: 246–250
7 *National Institute for Health and Clinical Excellence*. IPG149 Division of ankyloglossia (tongue-tie) for breastfeeding – guidance. NICE December 2005 ISBN 1-84629-121
8 *Madlon-Kay D*, *Ricke L*, *Baker J et al*. Case series of 148 tongue-tied newborn babies evaluated with the assessment tool for lingual frenulum function. Midwifery 2008; 24: 353–357
9 *Geddes D*, *Langton D*, *Gollow I et al*. Frenulotomy for breastfeeding infants with ankyloglossia: effect on milk removal and sucking mechanism as imaged by ultrasound. Pediatrics 2008; 122: 188–194
10 *Segal L*, *Stephenson R*, *Dawes M et al*. Prevalence, diagnosis and treatment of ankyloglossia, methodologic review. Can Fam Physician 2007; 53: 1027–1033
11 *Ricke LA*, *Baker NJ*, *Madlon-Kay DJ et al*. Newborn tongue-tie: prevalence and effect on breast-feeding. J Am Board Fam Pract 2005; 18: 1–7
12 *Ballard J*, *Auer C*, *Khoury J*. Ankyloglossia: assessment, incidence and effect of frenuloplasty on the breastfeeding dyad. Paediatrics 2002; 110: 1–6
13 *Kramer MS*, *Kakuma R*. Optimal duration of exclusive breastfeeding: a systematic Review. Cochrane Database Systematic Review 1: CD003517 2002
14 *WHO/UNICEF*. Global Strategy for Infant and Young Child Feeding. Geneva, World Health Organization; 2003
15 *Scientific Advisory Committee on Nutrition*. Infant Feeding Survey 2005. A commentary on infant feeding practices in the UK. The Stationary Office; 2008, ISBN 978-0-11-243118-3
16 *World Health Organization*. Protecting, promoting and supporting breastfeeding: the special role of maternity services. A joint WHO/UNICEF statement. Geneva, World Health Organization; 1989, ISBN 92-4-156130-0
17 *Morland-Schultz K*, *Hill PD*. Prevention of and therapies for nipple pain: a systematic review. J Obstet Gynecol Neonatal Nurs 2005; 34: 428–437