

Is tongue tie really the problem? Incidence of ankyloglossia in an infant population presented with suboptimal feeding: a cross-sectional survey

By Amy S. Miller, MSc¹ Joyce E. Miller, DC, PhD²

1. PhD student, Bournemouth University, UK

2. Associate Professor, AEEC University College, Bournemouth, UK

Corresponding author: Joyce E. Miller, DC, PhD

Email: jmiller@aecc.ac.uk

ABSTRACT

As the number of infants presenting to chiropractors with the problem of suboptimal breastfeeding is increasing, further questions about this population are raised. The incidence of ankyloglossia (tongue tie) diagnosis appears to be high in this population. However, there is little literature or clarity on the role of the ankyloglossia in the often complex clinical presentation of feeding difficulties, particularly on sustaining breastfeeding in the medium to long term. This study was designed to describe a population of infants presented to a chiropractic teaching clinic with the problem of suboptimal breastfeeding, and assess this population for diagnosis and management of ankyloglossia. This will be used as a starting point for further research into these common problems of ankyloglossia and suboptimal breastfeeding. Inclusion criteria were infants presenting to this clinic with the complaint of feeding difficulties, as reported by the mother. Data were collected using maternal questionnaires and a total of 131 infants were included over a period of five months. Ankyloglossia had been diagnosed prior to presentation to the chiropractor in 39% of infants, and of these, 77% had undergone frenulotomy (tongue tie cut) once or more. Given the high incidence of ankyloglossia diagnosis and frenulotomy in these infants with persistent feeding difficulties, both the diagnosis and management of this problem must be reflected upon and questioned. This study highlights a clinical need for a) clearer diagnostic criteria for ankyloglossia and, b) further research with a focus on sustained breastfeeding following frenulotomy and other treatments.

Keywords: Ankyloglossia; breastfeeding, chiropractic, frenulotomy, pediatric, suboptimal breastfeeding

Introduction

There is little consensus worldwide on the impact of ankyloglossia on breastfeeding difficulties, and the impact of frenulotomy on improving breastfeeding in these cases. While frenulotomy is encouraged in some countries, including the United States, it is not recommended in Canada or Japan, and has even been abandoned in the Netherlands.¹ Worldwide prevalence estimates vary greatly, partly due to a lack of definitive diagnostic criteria.¹ The current UK guidelines from the National Institute for Health and Clinical Excellence (NICE) state that there is little evidence for frenulotomy in improving the public health goal of higher long-term breastfeeding rates, and that conservative measures including massaging and exercising the frenulum should be implemented.²

There is growing evidence for the role of musculoskeletal dysfunctions contributing to suboptimal breastfeeding, and infants with suboptimal breastfeeding are increasingly presented to the chiropractor.³⁻⁶ It was noted in a previous study that the incidence of ankyloglossia (tongue tie) in a population presented for chiropractic care with suboptimal breastfeeding appeared to be disproportionately high at 58%,⁷ compared to worldwide estimates of 3-16%.⁸⁻¹¹ In cases of

suboptimal breastfeeding, musculoskeletal issues are also common,⁷ and it is anatomically plausible that ankyloglossia and musculoskeletal problems may coexist or even be directly associated.

It would seem perfectly reasonable to chiropractors, and indeed allied health care professionals, that an adult with tight hamstrings could have changes in posture and biomechanics of their low back. Similarly, a tight lingual frenulum has the potential to alter the biomechanics of the tongue, jaw and neck, and cause the infant to use suboptimal motor patterns in an attempt to suck.

Further, asymmetries of the head, jaw and neck are common in infants, and have been associated with birth trauma, birth intervention and prolonged second stage of labour.¹² This is reflected in the population of infants in this clinic presented with feeding difficulties, where a higher than expected rate of birth intervention was demonstrated.⁷ The importance of mobility of the tongue has been demonstrated in an ultrasound study, which described wedging of the anterior tongue between the nipple-areola and the lower lip, and undulation of the posterior tongue.¹³ This requires the infant to

be able to a) protrude the tongue as far as the lower lip and, b) have sufficient movement of the posterior tongue to create peristalsis and negative pressure.

There is variable interpretation of the role of ankyloglossia in these cases of suboptimal breastfeeding, making evidence informed management difficult. It has been reported that 50% of infants with ankyloglossia will not encounter feeding difficulties.¹ A systematic review has reported that there is moderate evidence for the effectiveness of frenulotomy.¹⁴ However, this study and many others only measure outcomes immediately after the frenulotomy has been performed, with no further follow up. This approach has been criticised for ignoring the potential of placebo effect.¹ Further it does not provide a wider public health context of sustained breastfeeding, for example amount of breastfeeding at six months, which is the recommendation of the World Health Organization.¹⁵ One web-based survey regarding the breastfeeding difficulties before and after frenulotomy found that 85% of the types of ankyloglossia were posterior.¹⁶ This study demonstrated post-frenulotomy reduction in presence and severity of nipple pain in all ankyloglossia classifications, and improved latch in posterior ankyloglossia.¹⁶ However, it is not clear how long after the procedure the survey was sent out, and relied on recall of pre- and post-frenulotomy breastfeeding difficulties at the time of the survey. A high incidence of posterior tongue tie (78%) was also identified in a prospective cohort study.¹⁷ This study used a breastfeeding self-efficacy scale and a visual analogue scale for nipple pain, and showed improvements in both of these measurements after laser frenulotomy.¹⁷

The evidence for frenulotomy affecting breastfeeding rates in the long term is unclear. Another systematic review states that frenulotomy appears to offer breastfeeding benefits in approximately 50% of cases.¹⁸ However this review only reports follow up breastfeeding outcomes from one study, and this was number of breastfeeds in a 24-hour period measured at two weeks after the intervention. Although the number of breastfeeds did increase in this study, this finding is without context and therefore does not necessarily support frenulotomy as a means to increase continued breastfeeding.

The lack of clarity in the literature is demonstrated in the variable approaches to the diagnosis and management of frenulotomy, and a new perspective is worthy of pursuit. Therefore, this study was designed to investigate a population with suboptimal breastfeeding for report of ankyloglossia, with a view to inform future study into this common and complex problem from a musculoskeletal perspective.

Methods and Material

This cross-sectional survey used data that was collected as part of routine clinical information provided by mothers at

initial presentation of their infant to a university-affiliated chiropractic teaching clinic. No recruitment techniques were utilized and the inclusion criteria was infants presenting to this chiropractic teaching clinic with feeding difficulties, as reported by the mother. Exclusion criteria was refusal for data to be used for research and inability to read English. The study period was October 2016 to February 2017, with a total of 131 infants presenting with feeding difficulties during this time. Questions included infant gender, age, feeding type and birth type. As part of a larger study, mothers also rated any difficulty feeding their infant on an 11-point scale, with 0 being no feeding problem, 5 being moderate problem and 10 classified as a serious problem. This question was validated during the development of the UK Infant Questionnaire,¹⁹ and was used at intake and discharge from the clinic. Data were summarised in Microsoft Excel. The survey was part of routine intake questionnaires at the clinic, which had been approved by the AECC Ethics Sub-Committee. All mothers included in the study signed consent for their infant's records to be used for research purposes on an anonymous basis.

Results

A total of 131 infants were included in this initial study. Table 1 shows the demographic profile of infants included. A limited number of cases had descriptive information about the feeding outcome following frenulotomy; seven mothers felt there had been no improvement or worsening in feeding after frenulotomy and two mothers felt it had made a significant improvement in feeding.

Table 1. Demographic profile of infants attending chiropractic clinic for breastfeeding problems

		n	%
Infant gender	Male	58	44%
	Female	73	56%
Infant age	Mean age infant	6 weeks	
	Mode age infant	1 week	
Feeding type	Totally breastfed	70	53%
	More breast milk than formula	38	29%
	More formula than breast milk	13	10%
	Totally formula fed	8	6%
	Other	2	2%
Birth type	Natural birth without interventions	50	38%
	Induced	43	33%
	Forceps	24	18%
	Ventouse	9	7%
	Emergency Caesarean section	23	18%
	Elective Caesarean section	9	7%

Figure 1 shows that at the time of initial presentation, 13 infants (10%) had not been assessed for ankyloglossia; 67 (51%) had been examined and did not have ankyloglossia, and 51 (39%) had been examined and diagnosed with ankyloglossia. Figure 2 shows that of the 51 infants who had a diagnosis

of ankyloglossia, 12 (23%) had not undergone frenulotomy, 30 (59%) had one frenulotomy, and 9 (18%) had two or more frenulotomies.

Figure 1. Diagnosis of ankyloglossia prior to attendance at breastfeeding clinic

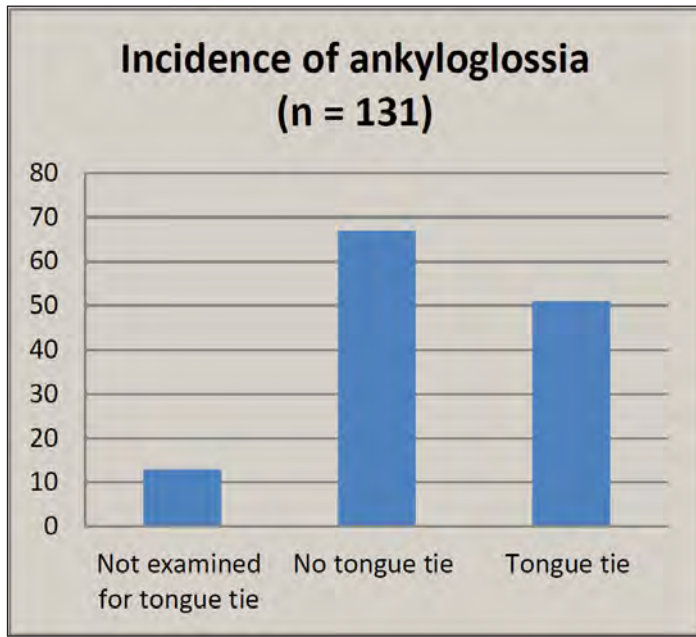
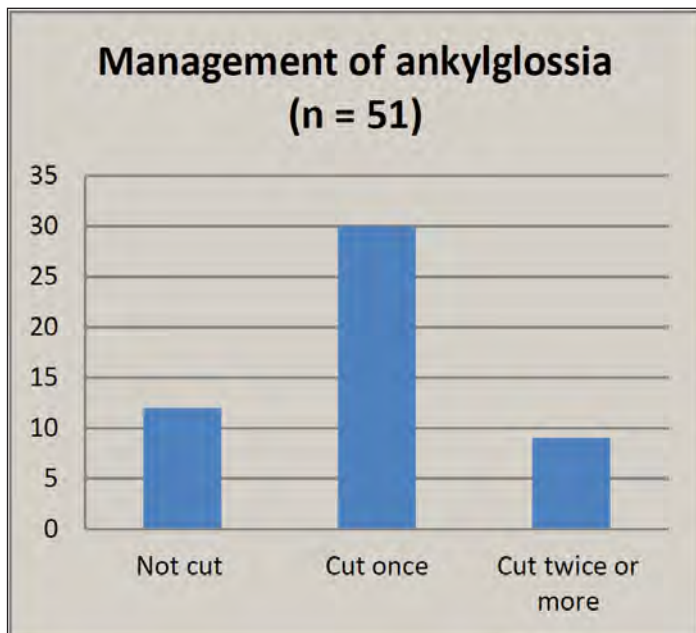


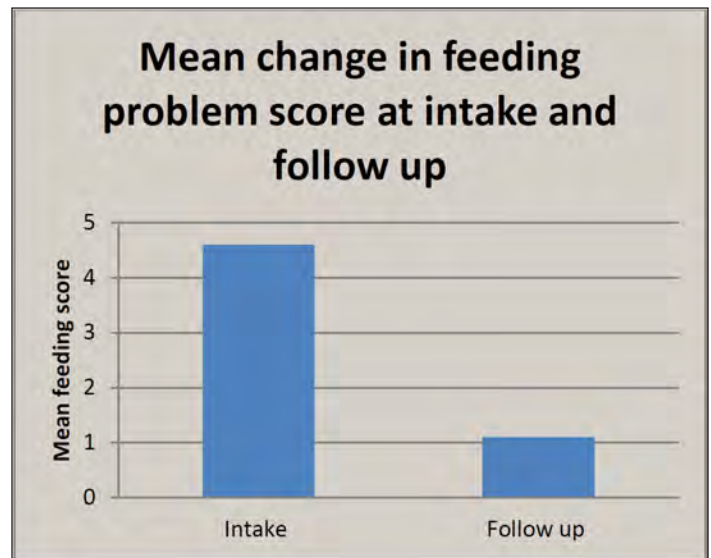
Figure 2. Management of ankyloglossia prior to presentation at the chiropractic teaching clinic



An incidental finding in this study was improvement in feeding with chiropractic management. At intake, the mean score of feeding difficulties on a scale from 0-10 was 4.6, this

reduced to 1.1 at follow up. This is shown in Figure 3. Follow up questionnaires were completed by mothers on discharge of the infant from the chiropractic clinic, after an average of four treatments. There was no further or longer term follow-up. The patients were discharged as in routine care when the mothers considered the problem to be resolved or no further improvement was expected. The average change (improvement) was 76%, which was statistically significant and also considered to be broadly clinically significant.²⁰

Figure 3. Change in maternal report of feeding problem after treatment in chiropractic clinic



Discussion

This was a survey of ankyloglossia in infants presented for the problem of suboptimal breastfeeding as part of routine intake information gathered in a chiropractic clinic. Prior to presentation at the chiropractic clinic, half of the infants had been examined and told there was no ankyloglossia. Just under half were diagnosed with ankyloglossia and these cases had undergone variable management prior to presentation at the chiropractic clinic. As these infants were later presented to the chiropractic clinic with feeding difficulties, it appears that previous management had not been successful in resolving the feeding in these cases. What is unknown is the quality of the treatment that was performed and one cannot discount the potential for a poorly performed surgical procedure.

There were four key issues highlighted in the results of this study.

1. Diagnosis: what constitutes a valid diagnosis of ankyloglossia?
2. Subgrouping: which babies diagnosed with ankyloglossia

should undergo frenulotomy, and which should not?

3. Management:

a. insufficient evidence for frenulotomy resolving breastfeeding difficulties, particularly in the medium and long term

b. insufficient evidence for other modalities in resolving breastfeeding difficulties associated with a diagnosis of ankyloglossia

The two main issues at the early stage of raising ankyloglossia as a potential problem are first, there are few standardised validated tools for diagnosing ankyloglossia. Second, there does not appear to be a validated decision-making tool to determine which infants with diagnosis of ankyloglossia will benefit from frenulotomy, and which will not.

There is a standardized tool, the assessment tool for lingual frenulum function (ATLAFF) developed by Hazelbaker.²¹ However, this tool has been shown to be too long and complex to use in a busy clinic, and further could not be used for more 60% of infants who were studied.⁹

A new instrument, the Bristol Tongue Assessment Tool (BTAT)²² has been developed with a view to solve those difficulties and to assist practitioners in tongue-tie identification. Their goal was to devise a four-step simplified procedure, but this has only recently become available for use and has not been validated.

It is apparent that despite best efforts at diagnosis and treatment, even frenulotomy does not always solve the problem, as cases presented to chiropractors had ongoing difficulty with feeding after the procedure. This raises the question as to whether frenulotomy is firstly, effective, and secondly, the appropriate management in all cases. Ankyloglossia may have been used as a “scapegoat” for the cause of the feeding problem, with other etiologies not sufficiently investigated. There is too little research investigating the role of frenulotomy in suboptimal breastfeeding, along with the quality of the procedure performed, ahead of any other types of intervention, on the effectiveness of frenulotomy on resolving breastfeeding difficulties, and on the long-term implications with regards to sustained breastfeeding. A recent systematic review reported that frenulotomy appears to offer long-term benefits to breastfeeding in a little over 50% of cases.¹⁴ NICE guidelines highlight that breastfeeding is a complex interaction between mother and infant, and that the evidence for frenulotomy in supporting breastfeeding is not of high quality.²

While frenulotomy is routine management once a diagnosis of ankyloglossia is made, there is little definitive evidence for long-term effects on breastfeeding. Of the babies diagnosed with ankyloglossia, more than three-quarters had undergone frenulotomy. Despite this, these infants were still presented for further care for the problem of suboptimal breastfeeding. There is insufficient research into the role of biomechanical care for the infant prior to or alongside frenulotomy in order to support and sustain the results of the procedure. The current guideline from the National Institute for Health and Care Excellence (NICE) for frenulotomy states that conservative measures for ankyloglossia include breastfeeding support, massage of the frenulum and exercising the tongue.² Chiropractic care has a role in providing musculoskeletal care and support for these infants. Some research has shown resolution of breastfeeding problems with chiropractic care.³⁻⁵ However, the care has not been delineated as to whether it assisted infants who had or had previously had ankyloglossia. Clearly, there is a role for future research that documents these types of cases, since they are so prevalent.

There are some interesting findings in the demographics. In previous studies of infants presented to chiropractors, there has been a slightly higher ratio of male infants to female.²³ Studies have also shown that there is a higher rate of ankyloglossia diagnosis in male infants.^{10,24} In this sample, this ratio was reversed, 56% were female and 44% were male.

The aims of this study were simply to describe this population of infants presented to a chiropractic clinic with breastfeeding difficulties, with a particular focus on ankyloglossia status. As such, this study provides more questions than answers. Further, the numbers are too small to define any indications beyond some possible trends. However, it does point out the great need for additional modalities of care for ankyloglossia beyond frenulotomy, and for research to be undertaken into those types of care.

Conclusions

Ankyloglossia was a common problem in this population with suboptimal breastfeeding. Further study to define a valid diagnosis of ankyloglossia, subgroups of suboptimal breastfeeding including ankyloglossia, and subsequent appropriate management, are essential. In addition, alternative or combined treatment options in the management of ankyloglossia should be investigated for effectiveness, including musculoskeletal care to address associated tissues and biomechanics.

References

1. Power RF, Murphy JF. Tongue-tie and frenotomy in infants with breastfeeding difficulties: achieving a balance. *Arch Dis Child* 2015; 100: 489-94.
2. National Institute for Health and Clinical Excellence. Division of ankyloglossia (tongue tie) for breastfeeding [NICE Guideline]. 2005. Available from: <https://www.nice.org.uk/guidance/ipg149/chapter/4-About-this-guidance> [Accessed 04 April 2017].
3. Vallone, S. Evaluation and treatment of breastfeeding difficulties associated with cervicocranial dysfunction: a chiropractic perspective. *J Clin Chiropr Ped* 2004; 6(1): 349-68.
4. Holleman AC, Nee J, Knaap SFC. Chiropractic management of breastfeeding difficulties: a case report. *J Chiro Med* 2011; 10(3): 199-203.
5. Miller J, Miller L, Sulesund AK, Yevtushenko A. Contribution of Chiropractic therapy to resolving suboptimal breastfeeding: a case series of 114. *J Manipulative Physiol Ther* 2009; 32(8): 670-4.
6. Vallone S. Chiropractic evaluation and treatment of musculoskeletal dysfunction in infants demonstrating difficulty breastfeeding. *J Clin Chiropr Ped* 2016; 15(3): 1301-5.
7. Miller A, Miller J, Taylor A, Way S. Demographic profile of 266 mother-infant dyads presenting to a multidisciplinary breastfeeding clinic: a descriptive study. [Accepted for publication, 2017].
8. Messner AH, Lalakea ML, Aby J, et al. Ankyloglossia: incidence and associated feeding difficulties. *Arch Otolaryngol Head Neck Surg* 2000; 126:36—9.
9. 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.
10. Ballard JL, Auer CE, Khoury JC. Ankyloglossia: assessment, incidence and effect of frenoplasty on the breastfeeding dyad. *Pediatrics* 2002; 110:e63. doi: 10.1542/peds.110.5.e63.
11. Ngercham S, Laohapensang M, Wongvisutdhi T et al. Lingual frenulum and effect on breastfeeding in Thai newborn infants. *Paediatr Int Child Health* 2013; 33:86-90.
12. Stellwagen L, Hubbard E, Chambers C, Lyons Jones K. Torticollis, facial asymmetry and plagiocephaly in normal newborns. *Arch Dis Child* 2008; 93:827-31.
13. Elad D, Kozlovskya P, Bluma O, Laine AF, Pob MJ, Botzerc E, Dollberg S, Zelicovic M, Sirae LB. Biomechanics of milk extraction during breast-feeding. *Proceedings of the National Academy of Sciences* 2014; 111(14): 5230-5.
14. Finigan V, Long AJ. The effectiveness of frenulotomy on infant-feeding outcomes: A systematic review. *Evid Based Midwifery* 2013; 11(2):40-5.
15. World Health Organisation. Breastfeeding [online]. Available from: <http://www.who.int/topics/breastfeeding/en/> [Accessed 12 June 2017].
16. O'Callahan C, Macary S, Clemente S. The effects of office-based frenotomy for anterior and posterior ankyloglossia on breastfeeding. *International Journal of Pediatric Otorhinolaryngology* 2013; 77(5):827-32.
17. Ghaheri BA, Cole M, Fausel SC, Chuop M, Mace JC. Breastfeeding Improvement Following Tongue-Tie and Lip-Tie Release: A Prospective Cohort Study. *The Laryngoscope* 2017; 127(5):1217-23.
18. Ito Y. Does frenotomy improve breastfeeding difficulties in infants with ankyloglossia. *Pediatr Int* 2014; 56:497-505.
19. Miller A, Huizinga B, Pinkster M, Telford ACJ, ten Heggeler JM, Miller JE. Development and testing of a multidimensional parent reported outcome measure for common presenting complaints of infancy: the UK infant questionnaire. *J Clin Chiropr Ped* 2016; 15(3): 1292-1300.
20. Hurst H and Bolton J Assessing the clinical significance of change scores recorded on subjective outcome measures. *Journal of Manipulative and Physiological Therapeutics*, 2004;27:26-35.
21. Hazelbaker A. The assessment tool for lingual frenulum function (ATLFF): use in a lactation consultant's private practice [thesis]. Pasadena, California: Pacific Oaks College; 1993.
22. Ingram J, Johnson D, Copeland M, Churchill C, Taylor H, Emond A. The development of a tongue assessment tool to assist with tongue-tie identification. *Arch Dis Child — Fetal Neonatal Ed* 2015; 100(4): doi: 10.1136/archdischild-2014-307503.
23. Miller J. Demographic survey of pediatric patients presenting to a chiropractic teaching clinic. *Chiropractic and Osteopathy* 2010; 18(33): doi: 10.1186/1746-1340-18-33.
24. Walsh J, Links A, Boss E, Tunkel D. Ankyloglossia and Lingual Frenotomy: National Trends in Inpatient Diagnosis and Management in the United States, 1997-2012. *Otolaryngology—Head and Neck Surgery* 2017; 156(4):735-40.