Critical Review: The Effectiveness of frenotomy in the treatment of breastfeeding difficulties in infants with ankyloglossia

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This critical review examines the effectiveness of frenotomy in the treatment of breastfeeding difficulties in infants with ankyloglossia. Study designs reviewed included: controlled clinical trials, cohort study, case series, observational study, and guidelines. Overall, research supports the effectiveness of frenotomy for treatment of breastfeeding difficulties in infants with ankyloglossia.

Introduction

Ankyloglossia, or tongue-tie, is a congenital oral abnormality, characterized by an abnormally short lingual frenulum (Wallace & Clarke, 2004). The Academy of Breastfeeding Medicine Protocol defines partial ankyloglossia as "the presence of a sublingual frenulum which changes the appearance and/or function of the infant's tongue because of its decreased length, lack of elasticity or attachment too distal beneath the tongue or too close to or onto the gingival ridge" (Ballard, J., Chantry, C., & Howard, C.R. 2004). Complete ankyloglossia in which there is extensive fusion of the tongue to the floor of the mouth is extremely rare (Amir et al., 2006; Ballard et al., 2004). Historically, tongue-ties were thought to affect breastfeeding and were commonly divided by frenotomy, an incision or 'snipping' of the frenulum, by the midwife at delivery (Griffiths, 2004; Wallace & Clarke, 2006). As formula milk gained popularity in the 20th century, the technique of tongue-division fell out of favour as most infants with ankyloglossia can bottle feed successfully (Griffiths, 2004). The majority of current medical and surgical opinion is that ankyloglossia rarely, if ever, causes feeding difficulties and therefore should not be divided (Wallace & Clarke, 2006). Although the clinical significance of ankyloglossia is controversial, many lactation consultants and some physicians believe that tongue-tie can cause breastfeeding difficulties; sore nipples, poor infant weight gain, neonatal hydration, and shortened breast-feeding duration have been reported to be possible consequences (Dollberg et al., 2006). Tongue-tie may prevent the infant from obtaining enough of the areola into its mouth to put adequate pressure on the lactiferous sinuses with a resultant poor stimulus for the milk ejection reflex leading to longer feeds and poor weight gain. The mother may also experience painful, bleeding nipples as the baby holds the nipple in the anterior mouth and compresses it between the upper and lower gums (Griffiths, 2004). Although many infants with ankyloglossia can adequately breastfeed, those that are experiencing feeding problems are not being referred for frenotomy, either due to failure of recognition of the cause, or current medical opinion preventing the appropriate referral (Wallace & Clarke, 2006).

With the increased popularity of breastfeeding in the last decade, there has been a resurgence of interest in frenotomy as a treatment for ankyloglossia and an exploration of the complications associated with the procedure in the modern era (Hogan et al., 2005; Griffiths, 2004). The use of frenotomy to treat ankyloglossia vary widely, however, there is a growing tendency among breastfeeding medicine specialists to favour the release of tongue-tie of the infant to facilitate breastfeeding and to protect the breast-feeding experience (Ballard et al., 2004). A review of the available literature is warranted to determine if frenotomy is an effective treatment for breastfeeding difficulties in infants with ankyloglossia.

Objectives

The primary objective of this paper is to critically evaluate the existing literature regarding the effectiveness of frenotomy on breastfeeding difficulties in infants with ankyloglossia. The secondary objective is to propose evidence-based practice recommendation regarding frenotomy on ankyloglossia in infants with breastfeeding difficulties.

Methods

Search Strategy

Computerized databases, including PubMed, Medline, and CINAHL, were searched using the following search strategy:
(ankyloglossia OR tongue tie) AND (breastfeeding OR feeding difficulties) AND (frenotomy OR tongue

division) AND (infants OR babies OR newborns OR neonates).

This yielded few results, therefore search was modified to:

(ankyloglossia OR tongue tie) AND (breastfeeding OR feeding difficulties) AND (infants OR babies OR newborns OR neonates).

The search was limited to articles written in English.

Selection Criteria

Studies selected for inclusion in this review paper were required to investigate the effects of frenotomy in the treatment of breastfeeding difficulties in infants with ankyloglossia. No limits were set on the demographics of research participants or outcome measures.

Data Collection

Results of the literature search yielded the following types of articles congruent with the aforementioned selection criteria: controlled clinical trials (2), cohort study (1), case series (2), observational study (1), and guidelines (1).

Results

Controlled Clinical Trials

Hogan et al. (2005) conducted a randomized, controlled trial on 57 tongue-tied infants with feeding problems to compare whether intensive support, advice and help from the lactation consultant or immediate division of the tongue tie works best and enables the infant to feed normally. The infants in the study were either breast-fed or bottle-fed. Within each of these groups, infants were randomized to a control group (behavioural treatment) or treatment group (immediate tongue division). In the breast-feeding control group, advice and help was given with positioning and attachment, and a plan of care was made with the mother. If this support and plan failed to produce any improvement after 48 hours, division was offered to these mothers. The bottle-feeding mothers were given advice on different teats and positioning when feeding. Once frenotomy was performed, the mother and baby were given help and support as needed with feeding immediately after division. The baby was allowed to feed for as long as he/she desired and then discharged. Telephone follow-up with the mother occurred at 24 hours, weekly for 4 weeks, and after 4 months to inquire about their progress, long-term results and any complications. Statistical differences between the groups were identified using Fischer's exact test for breast-and bottle-fed, separately and combined, on SPSS version 11. Results showed that of the 20 breast-fed controls, one improved, but 19 did not. At 48 hours, these 19 were offered division and all accepted. Of the 19 breast-fed babies who had immediate division, 19 improved and fed normally. Overall, division of the frenulum in the babies resulted in improved feeding in 54 out of 57 babies (95%). There were no problems with infection or bleeding, either primary or secondary. Most babies cried for only a few seconds until they were given a feed. The author concluded that division was safe and significantly improved feeding for mother and baby and division was significantly better than the intensive, skilled, professional support of the lactation consultant ($P < 0.001$).

The study was an open label study, and therefore experimenter and performance bias may have occurred. Subjects were randomized to groups which may control for any allocation bias. However, subjects in the control group were invited to the other arm of trial (group with frenotomy) after not responding to their arm of treatment, as it would be unethical to withhold potentially effective treatment. Hogan et al. (2005) reported detailed information on the subjects, however, inclusion and exclusion criteria were not discussed. Although the study stated the presenting symptoms of the breast-fed infants with ankyloglossia (latching problems, sore nipples, continuous feeds, top-up feeds), the authors did not report outcome measures for each symptom. Instead, the percentages of overall improvement in feedings for both groups were reported. Additionally, the authors reported following up of subjects, however, they did not report these specific outcome measures at these specific times. As well, the duration of behavioural treatment (24 hours) may have been too short to see any significant changes in the subjects. The authors used appropriate statistical tests for analyzing a relatively small sample size. Overall, this study offers significant evidence for the effectiveness of frenotomy in the treatment of breastfeeding difficulties in infants with ankyloglossia.

Dollberg et al. (2006) conducted a randomized, prospective, masked clinical trial of 25 infants with ankyloglossia and maternal nipple pain in which nipple pain and the infant's latch were compared before and after frenotomy in the alleviation of breastfeeding difficulties associated to ankyloglossia. Nipple pain and the infant's latch were measured through a standardized latch score and pain score

obtained from the mother by the lactation consultant. Results were compared using the Kruskal-Wallis test. In the 25 mothers, the authors reported a significant decrease in pain score after frenotomy than after sham and significant improvement in latch score after frenotomy. No significant side effects of the frenotomy were observed in any of the patients and bleeding (a few drops) was controlled within seconds in all cases. The authors conclude that frenotomy appears to alleviate nipple pain immediately after frenotomy and that it is effective in treating breastfeeding difficulties.

This study was well designed as it overcame several biases. It was a double blinded study which controlled for both participant and experimenter biases. As well, careful hemostasis (applying mild pressure) was completed after each frenotomy to ensure blindness. These are important strength to note as these biases were significant limitations in several other studies of frenotomy efficacy. The study was also randomized, therefore eliminating allocation bias. In addition, Dollberg et al. (2006) included descriptions of participants, recruitment procedures, and exclusion criteria. The statistical tests were appropriately used based on the non-parametric data collected in this study. In contrast to the strengths of this study, there were some limitations as well; the authors did not provide full details of frenotomy procedures or the validity of the measurement tools used, and there was no follow-up period to evaluate the long-term effects of the frenotomy procedure. Additionally, although the authors reported a significant change in both pain score ($P=0.001$) and improvement in latch score after frenotomy ($P=0.06$), they did not set criteria for statistical significance in their study. Standard specific practice often set a p-value of $p< 0.05$ as being "statistically significant" (Greenhalgh, 2006). Therefore, based on this standard value, the results of Dollberg et al. (2006) may have been inaccurate. Despite some methodological weaknesses, Doll berg et al. (2006) conducted a well controlled study and considered multiple variables. Their study provides strong evidence for the effectiveness of frenotomy in the treatment of breastfeeding difficulties in infants with ankyloglossia.

Cohort Study

Griffiths (2004) conducted a non-randomized, prospective cohort study to assess the indications for and safety outcome of, tongue tie division at one center of 215 infants younger than 3 months, who experienced difficulty breastfeeding. Feeding was assessed by the mothers immediately, at 24 hours, and 3 months after division based on maternal assessments. The authors reported that 80% were feeding better by maternal assessment at 24 hours and 57% noticed a difference immediately, and 64% breastfed for at least 3 months after division. 95% could poke out their tongues at 3 months. No anaesthetic or analgesic was used and there were no reports of significant complications.

This study was an open label study and therefore the health providers and subjects (mothers) were aware of the treatment given. This may have led to experimenter and performance bias as measurement outcomes were examined through maternal assessments regarding efficiency of the latch, the pain of chomping on the mother's nipple, or any improvement in the feeding and sleeping cycle. The sample size included was a reasonable size (one of the largest studies yet reported), however, there was no statistical analysis completed to determine whether a significant difference truly exists. A list of inclusion and exclusion criteria was used to select subjects and to ensure participants shared similar characteristics. Additionally, the inclusion of a control group in the study would have increased the study's validity. Although there was no control group to compare measurement outcomes, infants and mothers were all given help and support by their midwives, health visitors, infant feeding advisors, or lactation consultants first but were still unable to breastfeed. Given the multiple limitations of the study, it provides limited evidence of the effectiveness of frenotomy in the treatment of breastfeeding difficulties in infants with ankyloglossia.

Case Series

Ballard et al. (2002) examined 88 breastfeeding inpatient infants and 35 outpatient infants with ankyloglossia and with breastfeeding problems at a lactation center to define significant ankyloglossia, determine the incidence in breastfeeding infants, and measure the effectiveness of the frenuloplasty procedure with respect to solving specific breastfeeding problems in mother-infant dyads. After assessment for ankyloglossia and frenotomy was completed, mothers were asked to describe the sensation and quality of the sucks at the breast and then asked to grade her pain on a scale of 1 to 10. Pain levels were obtained after approximately 1 minute of latch on. Inpatients were followed for additional progress until discharge from the hospital and telephone contact was made routinely with each of the outpatients approximately 3 days after the procedure to ensure successful breastfeeding. Data were analyzed using SAS (SAS Institute Inc, Cary, NC). Univariate statistics were reported as mean+/-

standard deviation or as a percentage as appropriate. X^2 and Fisher exact tests were used for preliminary analysis of demographic variables comparing inpatient and outpatient subjects. Analysis was done using 2-sample and paired t test and McNemer's test as appropriate. Latch improved in all cases, and maternal nipple pain levels fell significantly after the procedure: 6.9 +/- 2.31 down to 1.2 +/- 1.52. There were no complications related to the procedure

Wallace & Clarke (2006) presented a case series of 11 infants who underwent tongue division for feeding difficulties and assessed the indications for and outcomes of the procedure. Only 10 of these 11 infants were followed up. Following tongue division, an improvement in breast feeding was noticed immediately by 4/10 mothers. Parents were subsequently contacted by phone by the first author at least 4 months after the procedure to enquire about the effect of the procedure on feeding and any complications encountered. Six out of 10 mothers successfully breast fed for at least 4 months. No anaesthetic or analgesia was used and there was little or no bleeding or infant distress. There were no reported complications of the procedure. This case series suggests a possible benefit of tongue division in symptomatic infants.

In the case series studies by Ballard et al. (2002) and Wallace & Clarke (2006), the authors did not provide a list of inclusion and exclusion criteria. In addition, results may have been affected by experimenter and performance bias. During the course of the study completed by Wallace & Clarke (2006), there was 1 subject lost to follow-up. Because sample size is considerably small, the inclusion of this subject may have altered the results of the study. Both studies did not include a control group, however, they did include pre and post treatment outcome measures. While Ballard et al. (2002) described the procedure for obtaining subjective outcome measures, the study by Wallace & Clarke (2006) did not provide any details regarding the measurement tools used. In addition, Ballard et al. (2002) did not use a quantitative measure of infant latch, relying only on their own observation and on the mother's description. As a result, it is difficult to assess the reliability of their ratings. Furthermore, Ballard et al. (2002) did not include a long-term follow-up of duration of breastfeeding and therefore, results may not be equated with "breastfeeding success". Finally, the authors of each study used appropriate analysis of their results based on their sample sizes. Although the study has a number of limitations, it does provide some guarded evidence of the effectiveness of frenotomy in the treatment of breastfeeding difficulties in infants with ankyloglossia.

Observational Study

Amir et al. (2005) conducted an observation study to review the first 12 months of assessment and release of lingual frenulum (frenotomy) at a breastfeeding clinic in a tertiary hospital of 66 infants and to report on the breast feeding outcomes and parental satisfaction. A structured interview was conducted by the mothers by telephone at least 3 months after the tongue –tie assessment by one of the lactation consultants. Data were collected about the presenting problem and the effect of the release of the tongue- tie on 46 infants. Parents were asked about their satisfaction with the procedure and about any problems following the release. Initial data collection was incomplete for 11 infants and nine were lost to follow-up. Follow-up data were collected on only 46 infants, and release was recommended and performed in 35 out of the 46 infants. After the tongue-tie release, 83% of mothers reported improvement in breastfeeding. Parents reported high levels of satisfaction with the frenotomy procedure and no complications were reported.

As an observational study, there are several limitations. The lack of control group to compare satisfaction outcomes, experimenter and performer bias, and the use of subjective measures may affect the strength of this study's evidence. However, it is important to note that this study served as a quality assurance project, and does provide support for the benefits of frenotomy in the treatment of breast-feeding difficulties in infants with ankyloglossia. This study also demonstrates the lack of complications associated with the procedure.

Guidelines

Ballard et al. (2004) developed a protocol to provide clinicians with guidelines for the evaluation and management of neonatal ankyloglossia and its complications in the breastfeeding dyad. The authors state that frenotomy may be considered appropriate for partial ankyloglossia and that it appears to be a minor procedure. However, frenotomy may be ineffective in solving the immediate clinical problem and may cause several complications, which however, are rare. Therefore, Ballard et al. (2004) recommends more research to be undertaken regarding benefits and risks of frenotomy for ankyloglossia and its effectiveness in treating breastfeeding concerns.

Discussion

Based on the available research, the literature demonstrates that frenotomy is effective in treating short-term breastfeeding difficulties in infants with ankyloglossia and that any complications associated to the procedure are rare. There is limited evidence, however, supporting long-term effects of frenotomy as treatment, as only the studies by Griffiths (2004), Hogan (2005), and Wallace & Clark (2004) included follow-up periods longer than 3 months. Therefore, assessments completed immediately after treatment, or within 24 hours, or even after 48 hours, may not be an adequate amount of time to determine the overall effectiveness of tongue division in treating breastfeeding difficulties. The study by Dollberg et al. (2006) is considered to be the only masked controlled study known to date, and based on critical appraisal, provides strong evidence for frenotomy as treatment for breastfeeding difficulties in infants with ankyloglossia. The other studies may serve as suggestive studies, based on their research designs and methodological limitations. It is also important to highlight that there is no consistency in the assessment tools used to diagnose ankyloglossia among the studies. For example, some studies based their assessments on observations gauged by eye, while others used a quantitative tool referred to as The Hazelbaker Assessment Tool for Lingual Frenulum Function (Ballard et al., 2002). In addition, there is a lack of consistency with outcome measures as each study measured different outcomes, most of which were subjective in nature.

Recommendations

The section above is an overview of the current available literature regarding the effectiveness of frenotomy in the treatment of breastfeeding difficulties in babies with ankyloglossia. A majority of the studies used research designs that were less rigorous than the gold standard of a randomized, controlled trial (Greenhalgh, 2006) and all of the studies demonstrate certain methodological weaknesses. Nevertheless, the studies collectively provide significant evidence for the effectiveness of frenotomy in the treatment of breastfeeding difficulties in infants with ankyloglossia. Therefore, it is recommended that frenotomy be considered an effective approach to treatment of breastfeeding difficulties in infants with ankyloglossia.

Further masked clinical controlled trials with longer time frames of assessment after the frenotomy procedure is completed are needed to determine the effectiveness of frenotomy in the treatment of breastfeeding difficulties in babies with ankyloglossia. Further research should also be established in the following areas to help clinicians maximize the effectiveness of frenotomy in the treatment of breastfeeding difficulties in babies with ankyloglossia:

1. Research to establish clinically practical and valid diagnostic criteria for ankyloglossia in infants
2. Research to develop a concise, practical, and standardized tool to assess ankyloglossia in infants
3. Research to establish objective outcome measures of frenotomy
4. Research to determine specific qualities of the frenotomy procedure (e.g. timing for procedure to occur after infant is born)

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