

Neonatal Tongue Tie: The Effectiveness of Intervention

Abstract

The promotion of breastfeeding is a major public health concern. The World Health Organisation and UNICEF express the importance of breastfeeding up to two years and beyond. Tongue tie is identified in the literature as abnormally short frenulum which can lead to breastfeeding difficulties such as attachment, nipple pain and damage. Tongue tie is viewed by the Baby friendly hospital initiative (BFHI) as a readily treatable cause for breastfeeding difficulties. This review aimed to identify if the intervention of frenotomy is effective in resolving breastfeeding difficulties. The themes that emerged from the comprehensive review of the literature were the prevalence and diagnosis of tongue tie, the impact of tongue tie on breastfeeding and treating tongue tie and its effect. Frenotomy is widely identified in the literature as successful in benefiting mothers and their infants to continue breastfeeding. There are some key recommendations made for practice in relation to tongue tie.

Review Article

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Abbreviations: DOH: Department of Health; WHO: World Health Organisation; UNICEF: United Nations Children's Fund; BFHI: Baby friendly hospital initiative; NICE: National Institute for Health and Care Excellence; CMS: Margaret Hynes Lactation Consultant

Introduction

The Global Strategy of Infant and Young Child feeding promotes the importance of breastfeeding from developmental, nutritional and immunological benefits (WHO-UNICEF [1]). Following the Department of Health (DOH) and World Health Organisation (WHO) / United Nations Children's Fund (UNICEF) recommendations, all hospital staff should encourage and enable mothers to breastfeed exclusively for the first six months and continue as part of a wider diet until two years of age or beyond. A global health campaign the Baby friendly hospital initiative (BFHI) has viewed tongue tie as a readily treatable cause for the failure to breastfeed. The National Institute for Health and Care Excellence (NICE) guideline on the division of tongue-tie for breastfeeding [2] define tongue tie as a congenital anomaly in which the baby is born with an abnormally short lingual frenulum. Griffiths [3] refers to the heart shaped characteristic the tongue may take in attempted protrusion because the edges of the tongue are curled up. Infants with a tongue tie can have difficulties with breastfeeding such as attachment, nipple pain and damage, poor infant weight gain and eventually leading to a reduction in milk supply. As breastfeeding benefits both the mother and infant the importance of addressing any condition that may impair feeding is a public health concern [4]. However, it must be considered that not all babies born with tongue tie experience feeding problems (UNICEF 2010). The Association of Tongue Tie practitioners (ATP) (2013) estimated about 25-60% of infants with tongue tie will present with feeding difficulties. The NICE guidelines [2] support the division of tongue tie (frenotomy) as an intervention, however it must be noted that this guideline is dated and further review and practice guidelines must be issued. The origins of frenotomy begin in Greek medicine where the practice was carried out for improved language and speech rather than feeding [5]. Aristotle described tongue tie as

'tongues that are slightly tied' leading to 'indistinct and lisping sounds'. With an increasing focus on breastfeeding and its benefits there has been a shift between frenotomy for speech improvement towards enabling better feeding. Becker [6] discusses the national rise in breastfeeding rates in 2010, when 58% of infants started their nutritional life with their mothers' milk.

The 2014 statistics from BFHI show that 61% of mothers initiated breastfeeding their babies. Bowley and Arul [7] do not recommend tongue tie release to prevent future possible speech and language difficulties, and refer to any significant interfering with feeding as the only indication for tongue tie release. Burrows [8] makes a valuable point regarding a women's motivation to breastfeed, how long she had intended feeding for and previous feeding experience all play a crucial role into successful feeding regardless of the presence of a tongue tie.

Aim

The aim of this literature review is to identify if the intervention of frenotomy for tongue tie was effective in resolving breastfeeding difficulties. The review critically appraises a number of research papers.

Methodology

The search strategy for this review was an exploration of medical and nursing databases including CINAHL, Medline, Web of Science, the Cochrane database, Science Direct and Ebsco host. The data reviewed was limited to that published between (2004 to 2016) and in the English language. Two search strategies were used in this review. The first search centred on tongue tie and breastfeeding and the second with a focus on tongue tie and frenotomy. 11 key studies were identified and used, further hand searches on key references lists were also carried out. The relevant Irish statistics came from the latest national perinatal statistics report from the ERSI. The Population Intervention Comparison and outcome framework (PICO) was used to extract evidence from systematic reviews, qualitative and quantitative studies (Appendix I).

Three core themes emerged from the review of the literature, the diagnosis/prevalence of tongue tie, 'the impact of tongue-tie on breastfeeding', and 'treating tongue-tie and its effect'.

The prevalence/diagnosis

According to Riche et al. [9] the lack of a consistent definition for tongue-tie has led to a wide discrepancy in reported prevalence of tongue tie. The reported incidence of tongue tie is variable with Hogan et al. [10] suggesting it could be as high as 1 in 10 infants with not all of these requiring treatments. Ballard et al. (2002) suggesting it affects 3.2% of infants. Lalakea et al. [11] reporting a prevalence of 0.02% to 4.8%, Ridgers et al. [12] suggesting 2.8%, Edmunds et al. [4] reporting an incidence of between 2.8% and 10.7% of infants. There is a suggested rise in the numbers of infants presenting with tongue tie. There are some questions regarding over diagnose now due to increasing breastfeeding rates. The ESRI perinatal statistics report [13] showed that 47% of mothers exclusively breastfed compared to 45% in 2009 and 42% in 2004. Similarly, Ritler [14] attributes the increasing rates with a mirrored increased breastfeeding rates. Ritler [14] denies any increasing prevalence of tongue ties suggesting babies have always been born it, suggesting when bottle feeding became the norm discussion around tongue tie reduced. Ritler [14] refers to the well know anecdotal tales that midwives used to keep one finger long and sharp to release tongue ties as a routine procedure.

The familial genetic inheritance of tongue tie is discussed by Klockers [15], In a case report Klockers [15] describes a Finnish family with isolated tongue tie as an autosomal dominant trait. In the article Klockers also refers to a dated study (1952) by Keizer involving a family of 26 members of 3 generations in which 13 had tongue tie inherited as an autosomal trait.

On review it must be taken into consideration that tongue ties are more common in male babies, Ridgers et al. [12] found a 64% male precedence, Argiris et al. [16] found 71% male and Hogan et al. (2005) found 61% male. However in a study by Mettias et al. [17] the male female ratio was identical at 49.2% and 50.8%. 'Tackling Tongue tie' was a campaign set up by the National Childbirth Trust (NCT) [18] in the United Kingdom due to an increase in numbers of parents contacting them with issues surrounding diagnosis and treatment of tongue tie.

In summary it can be concluded from the above evidence that there is undoubtedly an increased reporting of tongue-tie, and increasing awareness of tongue-tie as a cause of breastfeeding difficulties. However, there is a lack of certainty by researchers regarding the historic prevalence of tongue-tie in the general population, therefore it is impossible to say with any certainty whether prevalence of tongue-tie has truly increased, decreased or remained static.

O' Callahan et al. [19] suggest that the assessment for tongue tie should be an essential competency for midwives and Hughes [20] similarly suggests that there is no implication for incorporating tongue tie investigations into the existing neonatal examination. The author agrees that the assessment for tongue tie should be included in the initial examination of the new born but in order to insure consistency of diagnosis, the use of diagnostic tool such as the Hazelbaker tool (HATLEF) for presence of tongue tie

should be used. It also must be considered that the more health care professionals trained in assessment of tongue tie, may also increase the numbers of infants being diagnosed and referred for treatment. The presence of tongue tie does not always lead to breastfeeding problems (UNICEF 2010) (ATP 2013) (Hughes [20]) a factor which needs to be highlighted to professionals and women.

Amir et al. [21] conducted a quantitative assessment on the inter-rater reliability of the Hazelbaker Assessment tool for lingual frenulum function (HATLFF). 58 infants with tongue tie were referred to the breastfeeding education and support services at the Royal Women's Hospital in Melbourne Australia. The age range was 1 to 84 days with a median age of 10 days and 56% were male. There were also 25 infants in a control group assessed by two clinicians independently with an age range of 7-55 days and median age of 22 days. There was a 96% agreement between both assessors, there was no difference in the recommendations on tongue tie release. The study showed some discrepancies in agreement of infant sucking function but overall there was a high level of agreement on appearance of tongue tie assessment. The study concluded that the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) tool has a high reliability in the study of infants with tongue tie.

The impact of Tongue Ties on Breastfeeding

A phenomenological study by Edmunds et al. [4] carried out at a public health service feeding clinic in Queensland Australia to gain an understanding into the experiences of mothers who are breastfeeding an infant with tongue tie. Edmunds et al. [4] describes how the thickened, tightened or shortened frenulum in tongue tie affects the infant's ability to suck and often results in sore and painful nipples. The study received ethical approval from the University, the health service district and ethics committee. A Sample of 10 women was selected at their initial clinic visit when their infant was diagnosed with tongue tie. The data was collected over a 10 month period, 2 interviews were conducted, the first on the initial visit and the second two weeks later. 10 women whose infants were 3days to 3 weeks old participated in the study. 8 of these women were primgravidas, 7 had normal vaginal deliveries, 2 had ventouse assisted deliveries and 1 had an elective caesarean section. At the time of the second interview, 7 out of 10 infants had undergone frenotomy, the others choose not to and continued to breastfeed with some improvements and ongoing difficulties. The themes that emerged from the analysis were expectations, challenges, questioning seeking, perseverance and relief. The theme expectations describes the women's breastfeeding intentions, all 10 of the women intended to breastfeed and none had anticipated any problems. The 'challenges' theme describes how the women's breastfeeding experience unravelled when they ran into difficulties. One woman refers to the dread she felt before each feed due to the pain experienced.

"I ended up with cracked nipples, both sides ,both very sore... Sometimes it can take me half an hour to get the courage to put him on 'cause it hurts"

This is a clear indicator of how tongue tie can not only effect feeding but also change the way a women thinks about breastfeeding. The 'questioning' theme reflects how women began

to seek advice and help with their breastfeeding difficulties. A common trend amongst the women is about the conflicting advice they received from different staff in the hospital about feeding. For some of the infants tongue tie was identified in the hospital but no plan of care was put in place and it was rarely identified as the potential cause of breastfeeding problems. The theme 'perseverance' refers to how all of the women identified were determined to continue to feed despite the pain with the driving force behind the determination being their belief in the value of breastfeeding and importance of breastmilk. The 'relief' theme refers to the mothers whose infants had the frenotomy for tongue tie. The sense of relief is described by the women when tongue tie was identified as the probable cause of feeding difficulties and not as a result of anything they had done wrong. One woman describes how the frenotomy not only led to improved feeding but also allowed her to bond with her baby.

"It doesn't hurt like it did before, and it just feels normal, my nipples have started to heal, I think we are getting a better bond now because I am not scared of him." This study powerfully demonstrates what it is like for a mother to breastfeed an infant with tongue tie. A central finding of the study was the lack of knowledge healthcare professionals had about tongue tie and its effect on breastfeeding. It can be concluded for the women's words that tongue tie not only involves the physical difficulties of breastfeeding but can often also lead to an emotional trauma which can affect bonding. The key recommendations of the study are early identification of tongue tie and prompt and appropriate management including frenotomy.

A quantitative review by Todd and Hogan [22] undertaken at Canberra Hospital Australia, into the change in practice guidelines on tongue tie by comparison of patient characteristics and breastfeeding practices before and after the change. The change in guidelines recommended delaying tongue tie division until after 7 days of life. Data was collected on mothers and infants who had the tongue tie division in 2008 and those who had a division in 2011 after the new guidelines were implemented. The data collected includes gestational age, birth weight, gender age at time of division, degree of tongue tie, maternal issues around feeding with a tongue tie and the immediate post tongue tie division complications. Tongue ties were divided using the neonatal department guidelines, all were divided to the base of the tongue confirmed with a finger sweep to ensure complete division to the base of the tongue. The finding showed that there was no significant difference between the two groups in terms of gestational age, birth weight, and male to female ratio. The reported incidence went from 4.7% to 5%, the age at division increased appropriately according to the new guideline.

There was an increase 85.4 % to 97.2% in the mothers who had nipple pain when the procedure was delayed until after 7 days of life. These women were then more likely to bottle feed with either expressed breast milk or artificial milk due to the nipple pain. The significances to breastfeeding when a mother gives either expressed breast milk or artificial milk via bottle include a reduction in breastmilk supply, breast engorgement, mastitis and difficulty re-establishing the latch [22]. The study concluded that if feeding is problematic tongue tie should be divided as soon as possible to reduce early breastfeeding cessation. Berry et al. [23]

furthermore discuss the age of division, in Berry et al. [23] RCT the age range was 5-115 days, with the median age of 23 days, similar in other studies, Hogan et al. 2005 20 days median age, Argiria et al. [16] median age 4 weeks old. In studies, Ridgers et al. [12] discuss an early division undertaken at 10 days median age, Amir et al. [21] 12.5 days median age, the results concluded the same. The recommendations from Berry et al. [23], aim for division in symptomatic babies at 2 weeks of age in contrast to Todd and Hogan [22] who recommend that the division take place as early as possible. Todd and Hogan [22] suggest where a tongue tie is problematic an early division is recommended in order to establish and maintain breastfeeding as early as possible, reduce cessation and improve breastfeeding satisfaction. Conversely, Berry et al. [23] argues that a division too early can be met with the criticism that the baby may have feed well without the division.

A quantitative study by O' Callahan et al. [19], carried out in primary care practice in Connecticut in the United States explored the effects of office based frenotomy for anterior and posterior tongue tie on breastfeeding. The study recruited mothers of infants who underwent frenotomy from December 2006- through to March 2011, and then then completed an 18 question web based survey about breast feeding characteristics before and after the frenotomy. Information regarding the infant's date of birth, tongue tie classification, date of frenotomy and referral source were gathered on chart review. Research approval was granted by the Middlesex hospital Institutional review board.

Referrals for tongue tie were made from lactation consultants, physicians and craniosacral practitioners for breastfeeding difficulties or failure to gain weight, it must be noted that some infants also attended for a re-release of the frenulum, which had been carried out at a different facility but provided little to no improvement in breastfeeding. There were 311 infants evaluated and diagnosed with tongue tie, 299 underwent a frenotomy. 12 did not undergo the procedure due to parental choice. There were 157 mothers who responded to the survey. 98% of mothers had a breastfeeding consultation prior to being referred for frenotomy. The infant feeding latching difficulties pre and post intervention are discussed in the survey. 64% of the respondents who reported nipple pain during breastfeeding prior to the intervention reported no nipple pain one week post frenotomy procedure. Exclusive breastfeeding was reported by all 92% respondents post frenotomy with the mean duration being 14 months. 94% of participants reported no complications post the frenotomy. In the interpretation of these results it must be considered that 142 mothers (47%) did not respond to the survey, whilst those that responded had relatively positive experiences it must be questioned did those who did not respond have negative experiences therefore did not want to complete the survey. The study concludes that where tongue tie leads to maternal-infant feeding difficulties a frenotomy leads to improved breastfeeding characteristics. O' Callahan et al. [19] recommend that diagnosis of tongue tie should be a basic competency for all primary health care providers.

A quality review by Doyle and Barry [24] in a rural general hospital in Ireland, to assess why mothers stopped breastfeeding prior to discharge home from hospital. The review was carried out due to increasing rates of mothers who had initiated feeding

were not continuing to breastfeed on discharge 48-72 hours later. The review was carried out using data from one month period in 2013. The computerised Maternity Information system was used to identify all births and calculate the number of mothers who had initiated breastfeeding and stopped prior to discharge. Manual review of the charts to extract data in relation to demographic variables, antenatal factors, clinical issues, hospital practices, postnatal factors was carried out, as well as feedback given via service user comment form.

In the month reviewed, 102 babies were initially breastfed, 80 were discharged either exclusively or partially breastfeeding. 22 mothers that were initial breastfeeding their babies were no longer breastfeeding prior to discharge, these 22 mothers became the focus of the review. 7 mothers were primigravida, thus were breastfeeding for the first time, 15 mothers were multigravida, 11 of whom had breastfed before. 91% of infants in this audit were healthy term infants and 18% were in the special care baby unit (SCBU). The main challenges that emerged from the review were in relation to latching issues attributed to sore nipples as a result of tongue tie. The feedback from service users indicated that contributing factors to breastfeeding difficulties were staff shortages or staff not having the time to support breastfeeding. The recommendations of this review fails to address the area of tongue tie and are based around breastfeeding support in particular antenatal breastfeeding support. In summary, there is limited breastfeeding support available in the hospital and coupled with staff shortages women received little support in the early postnatal days. Although tongue tie is identified as leading cause of breastfeeding difficulties in this hospital there is no clear plan to identify tongue tie early or offer training to staff regarding identification and providing support to women.

Treating Tongue-Tie and its Effect

A randomised control trial by Berry et al. [23] conducted at Southampton General Hospital in the UK, where 60 breastfed babies with a diagnosis of tongue tie and feeding difficulties were randomly allocated into two groups: a group where division of the frenulum was carried out and a group where surgery was not performed. Infants were randomized to through a computer generated randomization. The parents were given written information regarding the study, where the tongue tie and feeding difficulties were confirmed and the procedure was explained. Written and informed consent was obtained by the parents. Both the parents and the observer were blind to the group, the ethics of this study must come into question although ethical approval was gained.

Although informed consent was gained the author questions how parents could have been truly blind to the procedure or lack of procedure taking place, which may result in some bias in the results. Prior to the procedure, a short feed was observed to assess feeding using the LATCH scoring system a combination of the latch, audible swallow, type of nipple, comfort, hold and the Infant breastfeeding assessment tool, a maternal pain scale (1-10) was also recorded. This assessment was used in order to add objectivity to what may have otherwise been a maternal subjective response.

The procedure was performed by separating the infant from the parents and wrapped securely using a towel, the tongue was divided using a sharp blunt ended sterile scissors and the floor of the mouth compressed with sterile gauze swab. The only difference between the procedures was whether the tongue tie was performed or not. The author questions how the parents would have really been blind to lack of procedure taking place. Berry et al. [23] outline that care was taken to ensure that there was no visual clues or unusual delays that might allow the mother or observer to be aware of the group the infant was in. It must be questioned how a mother would be unaware of a procedure taking place on her baby. The Association of Tongue Tie Practitioners (ATP) [25] issued an information sheet on the control of bleeding post tongue tie division, outlining that a small amount of bleeding post division is common and to be expected. Therefore the validity of these results must be questioned, as it may not be considered a true blind study. Following the procedure or non-procedure a follow up phone call was done on day one to determine any change in feeding and a subsequent phone call at 3 months again to record any feeding changes. The author acknowledges that whilst the initial contact timing was apt in order to assess immediate changes a more intense follow up may have achieved more informed findings. The initial results showed that a better latch was immediately achieved, reduced maternal pain and feeding more effectively were all key findings. The division group reporting a 78% improved feeding rate with the non-division reporting a 47% improved rate. Whilst Berry et al. [23] argue that these results show that the tongue tie release has an effect the author would argue the 31% who were in the non-division group and yet reported an immediate improvement in breastfeeding must be considered to have had a placebo effect.

The study whilst it demonstrates that frenotomy can be effective in alleviating breastfeeding difficulties, it is difficult to determine whether improvements in breastfeeding were due to the frenotomy procedure or intensive breast feeding support. An obvious limitation of this study is the brief and widely spaced follow up between assessments, which provide little information on prolonged breastfeeding success. The study fails to mention any links to the maternal motivation to breastfeed, previous breastfeeding experiences and cultural support which may have an impact on the longevity of breastfeeding. As mentioned previously by Berry et al. [23] these are crucial in the success of breastfeeding independent to the presence of tongue tie.

In a qualitative study by Amir et al. [21] which took place in a breast feeding clinic in a tertiary maternity hospital in Melbourne Australia, the infants were assessed using the Hazelbaker assessment (HATLFF tool) to assess the likelihood of tongue tie impacting negatively on breastfeeding. Amir et al. [21] utilised the tool as for diagnostics due to lack of agreed definition on what constitutes a problematic tongue tie, the tool includes five appearances such as length and attachment of frenulum, appearance of tongue when lifted, attachment of lingual frenulum to tongue and attachment of lingual frenulum to inferior alveolar ridge. If the infants were assessed as having impaired lingual function or the frenulum was visualised to be a thin membrane then a frenotomy was performed. Data was collected on 46 infants; at the time of assessment infants were median of 12.5

days old. The reported feeding problems were difficulty attaching baby to the breast, nipple pain, nipple damage, poor weight gain, frequent and prolonged feeding. After assessment of the frenulum with the Hatlff tool frenotomy was recommended in 76% of infants, frenotomy was performed. Following the procedure the infant was immediately offered the breast or bottle if appropriate. Four infants had the procedure performed in the ward, 28 in the feeding clinic immediately and three had it performed at a later visit. 51% of mothers noted better infant attachment to the breast, 57% reported less pain feeding and 17 % improved weight gain.

From this study the overall satisfaction rates were high and significant breastfeeding improvements are reported however the lack of hospital and ethical approval for the study as it was deemed unnecessary as it was used a quality assurance tool, make the results questionable.

Whilst many studies suggest that frenotomy is a painless procedure not requiring anaesthesia, McBride [26] made comparisons between frenotomy and circumcision and how it was similarly believed that that infants undergoing circumcision did not require anaesthesia. The protocol regarding the same was changed and McBride [26] questions whether the same would be done for frenotomy in years to come. An English Paediatric surgeon Fitz-Desorgher reviewed frenotomies undertaken in his clinic without anaesthesia between 1999-2001, there was a total of 144 infants. In 64 infants there was no bleeding, in 70 'a few

drops' of blood and 10 infants lost 'a small amount of blood'. Fitz-Desorgher concluded his review suggesting tongue tie division is easy, pain free, safe and usually successful. Ridgers et al. [12] suggests that infants are unlikely to find the procedure painful but may be uncomfortable and irritated by the finger inserted into their mouths. Burrows [8] acknowledge the few studies appraise the psychological impact of frenotomy on the infant or parent. The NICE guidelines [27] recommend that the use of anaesthesia is not required for infants under 3 months, undergoing a frenotomy [28-30].

Conclusion and Recommendations

The research under review demonstrates that the presence of tongue tie does impact on breastfeeding in most cases. The intervention of frenotomy emerges from the literature as the leading treatment for tongue tie. A significant gap and a limitation of the review was the lack of literature and limited research available from an Irish perspective. The author was concerned by this and is currently undertaking an audit in a maternity hospital in the Mid-West region of Ireland (Appendix II). The audit aims are to ascertain the effectiveness of intervention for tongue tie in Ireland. The audit will be carried out using a question based review and a follow up interview if required. The audit will be carried out and reviewed by the author, a Lactation consultant, a General Practitioner who is trained to carry out the procedure and a practice nurse.

Study	Patient problem	Intervention	Outcomes	Comparison
Ridgers et al. [12]	tongue tie	Frenotomy	Sample size 220	
London				
United Kingdom			64% male 141 Age at division (3-70) 67% feeding problems resolved 47% Improved feeding 5% no change to feeding	
Amir et al. [21]	tongue tie	Frenotomy		
Melbourne Australia			Sample size 66 Structured telephone interviews Age at diagnoses (3-98 days) median age 12.5 51% better attachment 57% improved sucking 17% improved weight	Improved feeding Age at diagnosis
Argiris et al. [16]			Sample size 46 median age 4 weeks Age at division 1 day to 12 weeks	
Chelmsford	tongue tie	Frenotomy	71% male 33	
United king			70% immediate improved feeding 78% improved latch 64% improved sucking	

Appendix 1: Data Extraction tool (PICO).

Baby Details:

Date of Birth : _____

Gestation Age : _____

Sex : _____

Birth Weight : _____

Mother :

Did you attend antenatal breastfeeding workshops? yes/no

Have you breastfed before? yes/no

If yes how long for ? _____

Did you receive breastfeeding support in hospital? yes/no

Was Tongue tie identified prior to hospital discharge?yes/no

Feeding difficulties identified prior to discharge:

- difficult to latch
- Sore nipples
- weight loss greater than 10 %
- Painful feeding

Who referred you to Dr. M? _____

Date of Procedure: _____

Did baby's feeding improve after procedure? yes/no

Did you continue to breastfed afterwards? yes/no

If yes, for how long did you continue feeding _____

Baby's Age now _____

Did you attend any complementary therapies? _____

If yes can you list some of the benefits? _____

Any further comments? _____

Thank you for taking the time to fill out this questionnaire.

Appendix 2: Tongue tie audit 2016.

The lack of a standard guidance regarding diagnosis, can lead to inconsistencies in intervention and the treatment used.

The author suggests that all healthcare professionals dealing with mothers and babies, midwives, public health nurses, paediatrics, G.P's and neonatal nurses be trained in the assessment of tongue tie and its impact on breastfeeding. The principle recommendation of this literature review is that a clear pathway be put into practice nationwide for the intervention and treatment once a diagnosis has been made. Further studies into pain relief during the procedure must be carried out.

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