All Tied Up! Influences of Oral Frenulae on Breastfeeding and their Recommended Management Strategies

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Background: *Recently, there has been an increased awareness of the role of the labial and lingual frenulae on a neonate's ability to latch and breastfeed efficiently. This critical review explores the (i) oral physiology of a baby nursing (ii) factors that can decrease a baby's ability to nurse efficiently, the problems these cause and their management and the (iii) relation between poor nursing efficacy and the risk of early childhood caries (ECC)* ***Study design:*** *An expansive search of the literature was performed using four electronic databases.* ***Results and conclusions:*** *Most studies assessing the role of labial and lingual frenulae on breastfeeding were of a low quality. The relation between ECC and poor nursing efficacy was found to be largely speculative. Hence, the results of these studies should be interpreted with caution. Despite the limited quality and external validity of the current evidence, in cases where breastfeeding difficulties are identified, surgical management of labial or lingual frenulae may provide some subjective improvements in breastfeeding outcome.*

Key words: *tongue tie, lip tie, ankyloglossia, frenotomy, breastfeeding*

INTRODUCTION

Breastfeeding is promoted as the ideal nursing practice for a newborn, due to the protective, immune-modulatory and nutritional advantages that breast milk confers plus the positive influence of breastfeeding on maternal attachment and mother-neonate bonding[1,2].

Despite the increased awareness and education around breastfeeding, the duration of breastfeeding is often short. Approximately, 25% of Australian mothers stopped breastfeeding by 6-months or earlier citing that their baby was not attaching properly while 18% reported that breastfeeding was too painful[3]. Recently, these concerns have been associated with the role of the labial and lingual frenulae and the baby's ability to attach and suckle effectively[4].

This critical review aims to discuss the (i) oral physiology of a baby nursing (ii) factors that can decrease a baby's ability to nurse efficiently, their subsequent problems, their management and, (iii) the relation between poor nursing efficacy and the risk of early childhood caries (ECC).

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METHOD

For consistency, 'nursing' is defined as the process of being fed at the breast[5].

Search Strategy

A literature search was completed in September 2016 using MEDLINE, CINAHL Plus, Web of Science and Academic Search Complete. No restrictions were placed on time period and all articles in English were searched. Additional hand searches of journals and reference lists of individual articles were completed. Keywords such as (nursing, breastfeeding, frenotomy, ankyloglossia) and MeSH terms such as "Breast Feeding" and "Dental caries" were used. Truncation (fren*, breast feed*) and ordered permutations of keywords were used to conduct a thorough search of the databases.

Although a systematic process was followed, the literature search was primarily expansive and based on a priori research questions with relevance studies being identified and discussed. To correspond with the primary aims of this review, the search strategy was structured into three parts.

The oral physiology of nursing

The oral physiology of a baby nursing centers around three main reflexes: rooting, suckling and swallowing[1,6-8].

The rooting reflex develops from 32 weeks' gestational age, when the baby's lip or cheek is stimulated, the baby gapes and turns towards the stimulus, attempting to grasp it orally.

The sucking reflex develops from 24 weeks' gestational age, and is stimulated when the nipple contacts the baby's palate. As the suckling process is initiated, tongue position becomes important to facilitate a seal between the oral structures and the breast. A seal is formed as the lips are turned outwards while the baby grasps the nipple and 2-3cm of the areola and the posterior portion of the tongue occludes against the soft palate.

The baby initially sucks rapidly until milk flow commences, the lateral borders of the tongue are elevated to form a trough through which the milk flows. Sucking also stimulates the synthesis and secretion of lactogenic hormones that evoke the mother's milk-production and ejection reflexes. As milk flow is established, a rhythmic peristaltic movement is initiated, moving milk from the anterior portion of the tongue down to the oropharynx. The suckling pattern is characterized by these deep rhythmic sucks in a suck-swallow-breathe cycle interspersed with periods of rest.

The swallowing reflex develops as early as 11 weeks' gestational age and is triggered as the volume of milk increases and begins to fill the oral cavity.

The coordination of these reflexes along with the baby's breathing begins between 32-35 weeks' gestational age. Notably by birth a healthy baby's breastfeeding reflexes are already well developed.

Mechanisms of breastfeeding

Two main theories have been used to explain how the infant extracts milk from the breast. The first suggests that compression of the breast by the infant's jaw coupled with the peristaltic action of the tongue allows for milk to be expressed[9]. While the second suggests that the infant creates an intra-oral vacuum during sucking that facilitates milk to be expressed[10]. Ultrasound studies suggest that milk begins to flow when the downward movement of the posterior tongue creates an intra-oral vacuum[7]. However, exact details of the suckling mechanism are still poorly understood and studies using real-time imaging and high image resolution are required[4].

The early postpartum period is a critical period within which the baby's instinctive actions and reflexes are consolidated to learned behaviours[1]. It is purposed that the use of other oral objects, such as teats, pacifiers and nasogastric tubes during the neonatal period may condition the infant to different oral actions thereby compromising the success of breastfeeding[1,11].

Factors that can decrease a baby's ability to nurse efficiently and their subsequent problems

Efficient breastfeeding relies on the complex interplay between the physiology and behavior of both the mother and the infant. Although the reflexes required for breastfeeding are already established at birth, the extraction of milk from the breast is an acquired skill that requires the baby to adequately latch and adjust his or her oral anatomical structures to facilitate adequate feeding.

There are several factors that have been described to decrease a baby's ability to feed efficiently; these can be broadly divided into:

1. congenital abnormalities: metabolic disorders, neurological disorders, syndromes, premature birth and conditions such as cleft lip and palate[1]
2. ankyloglossia/maxillary lip-tie[12,13]
3. oral dysfunctions: weak rooting and sucking reflexes, inverted lip position, biting patterns, excessive oral tension and altered tongue position[8]
4. miscellaneous: natal or neonatal teeth, use of orogastric or nasogastric tubes[14,15]

For the purposes of this review, a focus will be placed on the role of lingual and labial frenulae on breastfeeding.

Ankyloglossia

Ankyloglossia is a poorly defined anomaly but primarily involves a short, thick, fibrosed, or fixed lingual frenulum[16]. The prevalence of ankyloglossia ranges from 2-10% with a male predominance (2.6:1.0)[13,17]. A lack of consistency in the definition and classification accounts for a wide variation in the reported prevalence[13,17].

It has been postulated that ankyloglossia can cause a varying degree of reduced tongue mobility and functional limitations including: breastfeeding difficulties, atypical swallowing habits, speech articulation problems, mechanical problems such as inability to clean the oral cavity, and psychosocial stress[13,16,18].

The earliest discussions of ankyloglossia and its short- and long-term consequences date back to the early 2nd Century. Since then, the literature on ankyloglossia reflects the dominant views, influences and the evolution in management techniques, summarized in Table 1.

Despite the proposal of several classifications for ankyloglossia, to date, there is no universally accepted classification. A commonly used classification, described by Kotlow is based on measuring the 'free tongue' length as follows[16]:

Clinically acceptable, normal range of free tongue: >16mm
Class I: mild ankyloglossia: 12-16mm
Class II: moderate ankyloglossia: 8-11mm
Class III: severe ankyloglossia: 3-7mm
Class IV: complete ankyloglossia: <3mm

Sequelae of ankyloglossia

The sequelae are largely based on observational studies, case series and anecdotal reports, as listed in Table 2[16-21].

In ankyloglossia, the lingual frenulum tethers the tongue to the floor of the mouth, prohibiting its extension and movement beyond the lower gum. It has been suggested that this restriction prevents the infant from forming a good seal thereby resulting in insufficient extraction of milk from the mother[21,22]. Despite frequent feeds, the infant may have poor weight gain due to poor milk intake[7]. Furthermore, the abnormal and restricted tongue movements may cause nipple pain, bleeding, cracked or ulcerated nipples, mastitis and distortion of the nipple[20,23]. The prevalence of persistent nipple pain for mothers whose infant has ankyloglossia is between 36%-80%[24]. In nursing normal infants, it has been suggested that this pain is transient, peaks on the third day, and resolves spontaneously within 2 weeks with only 3% of mothers of normal infants have intractable pain or difficulty getting their babies to latch at 6 weeks[25]. Frequently, the level of discomfort and pain becomes unbearable thus leading to early cessation of breastfeeding[3]. Consequently, this can lead to maternal feelings of failure and negatively impact the psychosocial well-being of the mother[22]. In addition, ankyloglossia has also loosely been associated with mechanical problems related to oral clearance, patient-related psychological stress and orthodontic problems including malocclusion, open-bite tendency and spacing of the lower incisors[12,26] .

Impact of ankyloglossia on breastfeeding

Most studies evaluating the outcomes of surgical management of ankyloglossia have used objective assessment tools to gauge the degree of ankyloglossia and to evaluate breastfeeding difficulties. It is suggested that severity of these complications is determined by the degree of ankyloglossia[16].

Table 1: Impact of Ankyloglossia: A Historical Perspective

Year	Author	Location	Comments	Subject
350 BC	Aristotle	Stagira, Greece	*[in] the case of those whose tongues are slightly tied: their speech is indistinct and lisping*	Influence on speech
50 AD	Celsus	Rome	*In some the tongue is really attached to its base from the first day of life onwards, who therefore cannot speak…the tongue's tip should be seized with small forceps and the membrane thereunder incised But I have seen one who after cutting could protrude the tongue beyond the teeth, but still did not gain the ability to speak.*	Early description of frenotomy Influence on speech
200 AD	Galen	Pergamon, Greece	*Everything having to do with the tongue has been prepared most fully and perfectly by nature…nature has marvellously prepared a ligament of the size that would be most suitable… if it ended farther out along the tongue or came to an end sooner than it should, it would certainly not be as good for the articulation of the voice and it would also be a hindrance to the motion in chewing.*	Natural individual variations in frenum position
700 AD	Paulus Aegineta	Aegina, Greece	*Those who have the fault congenitally begin to speak late and have a tight band under the tongue*	Influence on speech
1584	Hieronymus Mercurialis	Forlì, Italy	*Not to tear off the membrane itself, but to perforate it near its root with a needle, and with this needle to pull through a thread, and to ligate it. This thread, tightened daily, will soon and gently detach the membrane.*	Frenotomy technique to reduce the risk of haemorrhage
1610	Jean Hérouard	Haute-ville-la-Guic-hard, France	*Seeing that he had trouble nursing we looked into his mouth. It was seen that the tongue-string was the cause. At five in the evening it was cut in three places by M. Guillemeau, the king's surgeon*	Impact of on nursing
1620	Hieronymus Fabricius	Acquapen-dente, Italy	*The great presumptuousness the midwives exert everywhere; when they without exception, disrupt the band under each new- born infant's tongue using their forefinger's nail, which they maintain sharp and pointed for this purpose; under the pretext that, if they wouldn't do it, the infant never would learn to speak*	Role of the midwife in frenotomy Influence on speech
1752	Justine Siegmundin	Germany	*For the need to loosen the tongue-band or membrane, these are the signs: That it cannot protrude the tongue or cannot move and wind it around the nipple. Among thousand infants there is barely one suffering from this defect*	Indications for frenotomy Prevalence
1791	Johann Hagen	Berlin, Germany	*Among ten infants, in whom the midwife or the nurse seek the reason for poor sucking in a short frenulum, barely one has this cause... Frequently the parents are deceived, for profit, greed and ignorance this aid is abused, and one unties where nothing is tied.*	Impact on nursing Controversies in management of
1906	Ernst Moro	Heidelberg, Germany	The anomaly rarely causes symptoms, but one can, to comply with the urgent wishes of mothers and midwives, cut the tongue band with a scissors kick to calm them.	Subjective improve-ments following frenotomy
1941	McEnery and Gaines	Chicago, United States of America	*In observing a large series of newborn babies, the authors have never seen a tongue that had to be clipped. Nursing technique does not require that the short frenum be cut and because of the dangers associ-ated with this so-called simple operation, they advise against it*	Indication for frenotomy
2000	Messner and Lalakea	Stanford, United States of America	*Most practicing lactation consultants believe that tongue-tie frequently causes feeding difficulties, and that neonatal frenotomy is quite helpful. In marked contrast, 90% of pediatricians and 70% of otolaryngologists believe ankylglossia never or rarely causes a feeding problem.*	Diversity of opinion in management
2007	Segal et al.	Montreal, Canada	There is no well-validated clinical method for establishing a diagnosis of ankyloglossia… Frenotomy is likely an effective treatment, but further randomized controlled trials are needed to confirm this.	Lack of validated classification Limited evidence base
2015	Francis et al.	Nashville, United States of America	*Frenotomy may be associated with mother-reported improvements in breastfeeding, and potentially in nipple pain…the strength of the evidence is low to insufficient.*	Subjective improve-ments following frenotomy Limited evidence base

Table 2: Reported Sequelae of Ankyloglossia

Infant Related	Maternal Related	Future Complications
Insufficient Extraction of Milk	Nipple Pain	Impaired Speech
• Poor Weight Gain	Nipple Trauma: Bleeding, Cracking, Ulceration	Malocclusion
• Failure to Thrive	Mastitis	Lower Incisor Spacing
Inefficient Extraction of Milk	Psychosocial Distress	Gingival Recession
• Poor Latch	Early Weaning	Difficulty Maintaining Good Oral Hygiene
• Poor Seal	Poor bonding between baby and mother	Increased Risk of Dental Caries
• Aerophagia		Difficulty Licking
• Reflux		Difficulty Playing Wind Instruments
• Irritability		Lower Self Esteem
Extended feeding time		

The Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) is commonly used to quantify the risk of ankyloglossia negatively impacting on breastfeeding[22]. Objective tools such as the LATCH (Latch, Audible swallowing, nipple Type, Comfort Hold) scores have been used to determine difficulties associated with breastfeeding thus identifying mothers at risk of early weaning[4,27,28]. Similarly, the Infant Breastfeeding Assessment Tool (IBFAT) facilitates the correlation between breastfeeding competence and maternal satisfaction[29,30].

Messner and Lalakea reported that 90% of pediatricians and 70% of otolaryngologists opined that ankyloglossia never or rarely causes breastfeeding problems[31]. Similarly, a recent systematic review emphasised that not all infants with ankyloglossia will have breastfeeding problems and many will adapt or respond to conservative therapy[13].

Posterior ankyloglossia

Posterior ankyloglossia and its association with the efficacy of breastfeeding has only been described by a few case reports17,32,33. Hong et al. described posterior ankyloglossia as a condition where the lingual frenulum is not very prominent on inspection but thought to be tight on manual palpation or found to be abnormally prominent, short, thick or fibrous cord-like with the use of a grooved director17. Posterior ankyloglossia remains a poorly defined condition and this is reflected by the significantly higher rate of revision frenotomies reported in these cases..

Management of ankyloglossia: Frenotomy, Frenuloplasty and Frenectomy

A frenotomy (also termed frenulotomy), is a minor surgical procedure involving separation or cutting of the frenulum, typically undertaken with sterile scissors, often without anaesthetic[18]. A more invasive frenectomy or frenuloplasty procedure may also be used, which combines excision and repair of tongue-tie, usually requiring the infant to undergo a general anaesthetic[18]. Frenectomy is reported to be a more predictable procedure resulting in a lower recurrence rate when compared to a frenotomy procedure[18,34].

Although most practicing lactation consultants recommend neonatal frenotomy to overcome the subsequent breastfeeding difficulties,[31] to date, there is no consensus on the indications and treatment options/techniques for management of ankyloglossia.

Frenotomy is a well-tolerated procedure and provides both objective and subjective benefits to the breastfeeding dyad[13]. Despite the poor quality of the current studies it appears that frenotomy/frenectomy procedures facilitates breastfeeding, enhances milk transfer to the infant and contributes to protection of the maternal nipple and overall breast health[13,35].

A recent systematic review[12] reported an improvement in breastfeeding effectiveness following a frenotomy procedure when compared with a sham procedure or no intervention[30,36,37]. However, the included studies in this review reported varying outcomes for frenotomy and decreasing maternal nipple pain[12].

There are no studies that have evaluated non-surgical interventions, and their long-term outcomes on breastfeeding and ankyloglossia[12]. However, the various surgical management techniques and treatment modalities that have been described, including their advantages and disadvantages are detailed in Table 3.

Maxillary lip-tie:

The attachment of the upper lip to the maxillary gingival tissue has been described by several terms including the superior labial frenum, median labial frenum and maxillary labial frenum[38]. If this attachment is tight, restricts the movement of the upper lip, or attaches near the maxillary alveolar crest, sometimes connecting with the incisive papilla, this is referred to as a maxillary or upper lip-tie. Kotlow's clinical classification described lip-ties based on the position and attachment of the frenum along the maxillary ridge[38].

Sequelae of maxillary lip-tie

It has been postulated that a tightly attached maxillary labial frenum may interfere with lip flanging resulting in a poorer latch[38-40]. However, there is a paucity of high quality evidence that explores the relationship between the maxillary labial frenum and breastfeeding outcomes.

Case reports have suggested that the presence of an maxillary lip-tie may be associated with prolonged and frequent breastfeeding with poor milk transfer, inability to maintain an effective latch, colic and reflux-like symptoms[19]. Furthermore, mothers may develop plugged ducts, pain during feeding, flattened, compressed and injured nipples, and mastitis[19]. Subsequently, this may lead to poor suck, poor seal around the breast, loud clicking noises due to tongue recoil, extended feeding times, poor infant weight gain, irritability and even infant reflux and aerophagia[19].

Table 3: Management Options for Ankyloglossia and Upper Lip-tie

Technique	Description	Advantages	Disadvantages
Frenotomy	Simple release of the frenulum	Rapid Relatively easy procedure Minimal equipment With or without local anaesthesia Can be performed in the neonatal nursery or clinic Can be breastfed immediately after the procedure	Risk of haemorrhage Need for clinical stabilisation Higher recurrence rate
Frenuloplasty/ Frenulectomy	Complete release of the frenum with plastic closure	Reduced scarring Lower recurrence rate	Requires a general anaesthetic for neonates Technique sensitive More involved than a frenotomy Longer procedure
Modality	**Description**	**Advantages**	**Disadvantages**
Scalpel or sterile scissors	Haemostat used to clamp the depth of the vestibule. Small sterile scissors/scalpel is used to release the frenulum, beginning at its free border and proceeding posteriorly directly adjacent to the tongue	Relatively quick and simple Can be achieved with no/topical anaesthetic Infant can be breastfed immediately which will also indirectly act as a tamponade to control bleeding	Multiple cuts may be required for visualisation/retraction and to achieve adequate release May require post-operative analgesia May require sutures Longer treatment time compared to other modalities
Laser	CO2 Laser (10600 nm and output power of 1.5 W, frequency of 100 Hz and pulse duration of 400 µsec) Erbium: Yag Laser (2970 nm, laser energy of 20 Hz and 65 mJ, 600µsec pulse duration) Diode Laser	Very quick Minimal bleeding Can be achieved with no/topical anaesthetic Reduced scarring Low risk of infection (wound sterilisation) Low post-operative pain (cauterisation of nerve endings)	Cost of equipment Laser safety and additional precautions
Electrocautery	Incisions placed using a needle electrode, muscle fibers separated using a loop electrode, coagulation achieved using a ball electrode	Relatively quick and simple Minimal bleeding	Requires adequate anesthesia Young children may require a general anaesthetic Risk of burns

Frenotomy, frenuloplasty and frenectomy are techniques that have been advocated in infants with a maxillary lip-tie encountering difficulties in breastfeeding[19,38]. Case reports also suggest that surgical release of the lip-tie improves the infant's ability to flange and latch. However, without appropriate controls, adequate samples and long-term follow-up, the results of these reports remain debatable[38].

Breastfeeding efficacy: a multi-factorial issue

Impaired tongue function results from the interaction of infant-related and maternal-related factors including pyschosocial and cultural behaviors patterned on the intricate mother-infant relationship[41]. Pransky and colleagues reported that many newborns (n = 127, 21%) who were referred with a suspicion of oral cavity anomalies causing difficulties in breastfeeding, were deemed not to have any oral cavity anomalies[33]. This highlights the notion that there are multiple reasons why a newborn may have difficulties in breastfeeding and a thorough diagnosis is critical to facilitate appropriate and timely interventions[33].

Possible relation between poor nursing efficacy and the risk of ECC

ECC is the presence of 1 or more decayed (noncavitated or cavitated lesions), missing (due to caries), or filled tooth surfaces in any primary tooth in a child 71 months of age or younger[42]. The etiology of ECC is multi-factorial and involves the complex interplay between dietary carbohydrates, cariogenic bacteria and dental hard tissues[43,44].

The carbohydrate content of breast milk or formula along with factors such as contact time with the tooth surface is attributed to the increased caries risk of an infant[45]. This risk is accentuated with the frequency of feeding and feeding practices which result in pooling of breast milk or formula around the teeth, for example nocturnal feeding[45]. Therefore, feeding habits have a pivotal role in the development of carious lesions[46].

Kotlow proposes that although breastfeeding alone may not cause dental caries, breastfeeding in conjunction with a maxillary lip-tie may be an underlying factor to caries development[47]. A maxillary lip-tie may create pockets allowing milk to pool, thus creating a cariogenic environment resulting in the development of dental caries on the labial surfaces of the infant's maxillary incisors[47].

Similarly, infants with ankyloglossia may take a longer time to feed and require more frequent feeds throughout the day, which is likely due to ineffective latching and suckling[48]. Therefore, it is logical to suggest that the increased frequency of milk and contact time against the tooth surface will increase the infant's caries risk[44]. Furthermore, the restricted tongue movement may reduce the ability of food debris to be cleared from the mouth and thereby predisposes the infant to dental caries[49]. Despite these hypotheses, there remains no high-quality evidence to support a correlation between ankyloglossia or maxillary lip-tie and an increased risk of ECC.

Bottle-feeding may be an appealing option for mothers who encounter breastfeeding difficulties particularly with an infant with ankyloglossia or a maxillary lip-tie. Ricke et al. suggest that bottle-feeding is a more passive process that poses fewer issues in cases of ankyloglossia[21]. If an infant is unsettled or irritable due to inefficient feeds, nocturnal or on-demand bottle-feeding may be introduced. Additionally, fermentable carbohydrates, a sweetened pacifier or snacks may be offered to settle the child. These changes in dietary practices may greatly increase the infant's caries risk predisposing to the development of carious lesions[50]. To date, there is a scarcity of studies evaluating the relation between the above-mentioned factors and risk of ECC, therefore further studies investigating these factors are essential to make valid correlations.

CONCLUSIONS

In light of the current evidence, one can conclude that the:

- oral physiology of breastfeeding revolves around three key reflexes that are essential in achieving good latch, seal and facilitating the expression of milk
- efficient breastfeeding relies on the complex interplay between the physiology and behavior of both the mother and the infant
- correlation between poor nursing efficacy and ECC is largely hypothetical with limited supporting evidence
- current body of literature, albeit of limited quality and external validity, tends to suggest that in cases where breastfeeding difficulties are identified, the surgical management of ankyloglossia or maxillary lip-tie may provide only subjective improvements in breastfeeding outcomes. Nevertheless, this requires cautious interpretation, as most studies lack a comparison or control group and are limited by factors such as selection and observer bias, subjective measures of assessment and limited follow-up times.

Therefore, future research endeavors would benefit from standardized methods of diagnosing and classifying ankyloglossia and maxillary lip-ties with the collection of additional longitudinal data allowing for both comparative and temporal analysis.

In the words of Celsus, from the first century AD: *"In medicine it may be universally valid what should be done, but not universally valid what results and whom it helps."*

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