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Review

Lingual frenotomy in neonates: past, present, and future

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Abstract

During the last decade, increasing awareness of breastfeeding and its health benefits has not been reflected in the provision of lingual frenotomy in neonates with tongue-tie. This could be because of inconsistencies in our understanding of the importance and treatment of ankyloglossia. In this review, we discuss the current clinical guidance on diagnosis and management, and the future of such a service in the early postpartum period.

Keywords: Lingual frenotomy; tongue-tie; neonates

Introduction

Almost 2000 years ago, Cornelius Celsus, a Roman encyclopaedist at the time of Christ, described the condition known as tongue-tie, and accurately noted the dangers of frenulotomy to release the tongue.[1] In the Bible, Moses complained, "I am slow of speech and of tongue" (King James version, Exodus 4:10), and St Mark wrote, "The string of his tongue was loosed, and he spake plain" (King James version, Mark 7: 35).

The midwives at the time of Fabrizio d'Aquapendente (a 16th century anatomist and surgeon, known in medical science as the "Father of Embryology") kept a fingernail sharp to strip the tongue of all new-born babies,[1] and the practice was also noted in John Theobald's *A Young Wife's Guide to the Management of Her Children* (London, 1764).[1] In the early 1900s, tongue-ties were routinely divided but, as formula milk gained popularity and infants could bottle feed, the practice fell out of favour.[2]

More recently, leading authorities, including the World Health Organization (WHO), the National Institute for Health and Care Excellence (NICE), and the Department of Health,[3] have recommended exclusive breastfeeding for six months postpartum,[4] and this has renewed interest in ankyloglossia and its effect on feeding. The increase in demand for treatment, however, has not been reflected by provision within the National Health Service (NHS), and mothers' concerns regarding the "postcode lottery" have attracted media attention.[5]

The treatment and importance of neonatal ankyloglossia have been controversial. We discuss the current guidelines in accordance with the best evidence available, and the future provision of treatment in the early postpartum period.

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Definition

Ankyloglossia is derived from the Greek words "agkilos" meaning curved and "glossa" meaning tongue. There is no universally agreed definition. In the 1960s, Wallace defined it as "a condition in which the tip of the tongue cannot be protruded beyond the lower incisor teeth because of short frenulum linguae, often containing scar tissue."[6] Elsewhere it has been defined as "the condition in which the tongue cannot make contact with the hard palate or cannot protrude more than 1-2 mm past the mandibular incisors", whilst the Academy of Breastfeeding Medicine defines it as 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".[7] It is universally accepted as a condition in which there is a short, tight, lingual frenulum that may be associated with a bifid tongue.[8]

Embryology

The tongue develops during the fourth to seventh weeks of the embryonic stage of pregnancy, when it is fused to the floor of the mouth.[9] During this period, the lingual frenulum serves as a guide for the forward growth of the tongue. The cells of the frenulum later undergo apoptosis, and ectodermal tissue, which surrounds the tongue, grows downwards. The tongue then separates from the floor of the mouth, increasing its mobility. Incompletion of this process results in ankyloglossia.

Pathophysiology

The exact pathophysiology of this congenital abnormality is unknown.[10] The mucosa covering the anterior two thirds of the mobile tongue is derived from the first pharyngeal arch, and deviation from its normal development is the most likely cause of abnormalities in the length and attachment of the frenulum.

Most cases are seen as a sporadic isolated finding in an otherwise normal child, but there can be a strong family history.[11] Familial patterns of ankyloglossia with autosomal dominant, autosomal recessive, and X-linked inheritance, have been reported in small case series.[12] No specific genes have been associated with isolated cases, but mutations in the T-box transcription factor (TBX22) gene on chromosome X have been linked to the condition with non-syndromic cleft palate.[13,14] TBX22 is the important susceptibility gene that is currently found in X-linked cleft palate – ankyloglossia syndrome (CPX), and it is an important genetic determinant for familial cleft palate, particularly when combined with ankyloglossia.

Several other syndromes are also associated with the clinical findings of ankyloglossia, including Ehlers-Danlos syndrome, Beckwith-Wiedemann syndrome, Simosa craniofacial syndrome, orofaciodigital syndrome, van der Woude syndrome, Kindler syndrome, and Optiz syndrome.[11,15,16]

Interestingly, in their study of the risk factors of ankyloglossia in 500 neonates, Harris et al showed that morbidity was more than three times higher among infants whose mothers smoked cocaine than it was among infants whose mothers did not, possibly as function of lower rates of mitosis.[17]

Development of swallowing

Swallowing can occur in utero from around 11 weeks of gestation. True sucking begins from weeks 18 to 24 and is characterised by a distinct backward and forward movement of the tongue. The link between sucking and swallowing is usually well established by 32 weeks of gestation. At 34 weeks, a healthy preterm infant is likely to suck and swallow well enough to sustain nutrition strictly through oral feeding, with backward movement more pronounced than forward movement.

Mechanism of swallowing

The movement of the tongue during feeding in infants has been studied by cineradiography and more recently by ultrasound.[18,19] Ultrasound shows some similarities between the movements babies make when both breast and bottle feeding. The tongue, however, is protruded further forward when breastfeeding, and the human nipple elongates during each suck, which an artificial teat cannot do.

Ultrasound studies have shown that the pattern of movements together with an intraoral vacuum, are important for the effective removal of milk from the breast.[19]

The swallowing mechanism of a newborn and infant is different from that of an adult or older child. To feed successfully, an infant must latch on to their mother's areola with the ridge of their upper gum, buccal fat pads, and tongue. The body of the tongue then forms a groove to pull the breast into the mouth and stimulate the release of milk. The tongue remains grooved and, with its body raised, the posterior aspect is lowered and the groove creates negative pressure to release the fluid. The tongue maintains a biphasic pattern during sucking, the jaw opens slightly while the anterior tongue and lip maintain a seal, and the anterior and upward peristaltic movement of the posterior tongue triggers the swallowing action to the pharyngeal wall.[12,19] During coordinated feeding, the movements of sucking, swallowing, and breathing, follow in a 1:1:1 sequence.

Immunity of neonates and its impact on virulent microflora in the mouth and gut

Pregnancy

It was previously thought that the in-utero environment was largely sterile and that a fetus was not colonised with bacteria until the time of birth. Recent studies, however, suggest that colonisation begins well before delivery.[20] In neonates, the gut microbiota develops by the third trimester of pregnancy when the fetus swallows nutrients, growth factors, and antibiotic, peptide-rich, amniotic fluid, which is a complex that is essential for its well-being.

Breastfeeding

Human milk is even more complex than amniotic fluid. The neonate's immunity continues with breastfeeding as the mother's milk improves intestinal development after birth. In addition to its nutritional composition, human milk contains hormones, growth factors, cytokine immunomodulators, natural peptides, antibiotics, sIgA, and probiotic bacteria.

Nutrition has an important role in the early colonisation patterns of gut microbiota in neonates. The difference in colonisation and transmission of immune-modulating factors between breast and formula-fed infants may have far-reaching effects over the course of a person's life, as it may have an impact on health.

Bottle feeding

The carbohydrates, bacteria, and nutrients that are found in formula-fed infants form different patterns of colonisation with different immunomodulatory effects on their developing gut microbiota. Whether or not formulas are milk-based or soy-based, the gut microbiota compromises a diverse array of bacteria, including *Escherichia coli*, *Clostridium difficile*, Bacteroides spp, Prevotella spp, and Lactobacillus spp.[20]

Epidemiology

Published papers about ankyloglossia consist largely of case series and observational studies. Reported incidence varies between 0.1% and 10.7% depending on the population studied and the criteria used to define the condition.[10,21] The variation may be attributed in part to the lack of a uniform definition and objective grading system.[22]

The incidence is higher in neonates than in children and adults,[6] and most studies have shown that it is more common in males.[2,8,10,11,17,23,24] Some studies show a higher incidence in European and English newborns,[25] whilst others report no ethnic predilection.[10,17]

Table 1
Criteria for the diagnosis of ankyloglossia.[21]

Clinical findings and criteria
Length of frenulum:
Extending along 25%-100% of tongue's total length
Abnormally short
< 1 cm long
Thickness of frenulum:
Thick
Thick & fibrous
Extension of frenulum:
Extends to the papillated surface of tongue
Clinical appearance of frenulum on protrusion of tongue
Tongue heart-shaped
Tongue displays notching
Tongue prevented from protrusion
Tongue tip fissures on normal movement
Mobility of tongue:
Reduced
Tongue tip cannot swing from one corner of mouth to the other
Tongue cannot reach gum when protruded

Diagnosis

Various classifications of ankyloglossia have been proposed but none has been uniformly accepted. It is often defined in terms of function based on neonatal oral anatomy, rather than on the basis of objective anatomical measurement. Currently, we know of no accepted standard definition, grading system, or practical guide on which to base diagnosis.[10,11,21]

Clinical presentation of ankyloglossia

The clinical phenotype varies from an absence of symptoms to rare cases of complete ankyloglossia in which the tongue is fused to the floor of the mouth. A commonly used criterion was that of the frenulum being abnormally short and thick, which caused the tongue to become heart-shaped on protrusion. Other functional impairments include reduced mobility of the tongue and an inability to protrude the tongue past the gum line.[26] Most surgeons routinely assess the extent of tongue-tie by observing the tongue's mobility on protrusion.[27]

Table 1 shows the diagnostic criteria listed in a review by Segal et al.[21]

Assessment tools

Currently, we know of three published objective measurement tools to assess the extent of the tongue-tie.

Table 2
Kotlow's classification.

Type of ankyloglossia	Movement of the tongue (mm)
Clinically acceptable (normal range of tongue movement)	> 16
Class I (mild)	12 - 16
Class II (moderate)	8 – 11
Class III (severe)	3 – 7
Class IV (complete)	< 3

The Hazelbaker assessment tool for lingual frenulum function (1993)

This quantitative instrument, which is used to assess the potential impact of a tongue-tie on breastfeeding, includes seven items on appearance and five on function.[28] Although it is comprehensive and can reliably indicate the need for frenotomy,[7] it is not simple to use in a busy hospital setting.[29]

There also seems to be a lack of agreement in terms of function score, and Madlon-Kay et al found that the scores of many babies did not fall into any of the three categories.[30]

Kotlow's classification (1999)

This classification was designed to assess the range of tongue movement (Table 2).[31]

The Bristol tongue assessment tool (2015)

This is based on clinical practice with reference to Hazelbaker, and was introduced as a simplified objective measure of the severity of tongue function (Table 3).[32]

Symptoms of neonatal ankyloglossia

Many neonates with ankyloglossia will be asymptomatic, but if left untreated, symptomatic cases can cause functional difficulties with breastfeeding, atypical swallowing, problems with deglutition, and poor attachment. It can also result in infants being unsettled, failing to gain weight,[33] and dehydration.[34]

In 25%–60% of infants it is associated with failure to thrive and refusal of the breast, and in mothers, painful and damaged nipples[4,35]and a poor milk supply. There is a 10% - 26% risk that breastfeeding will cease during the initial three weeks.[36]

The recommendation by the World Health Organization that mothers should breastfeed exclusively for the first six months and, with supplemental feeding, continue to do so until the age of 2 years, has been adopted in the UK because of its benefits to health.

Management

Ankyloglossia can present as an asymptomatic condition and may resolve spontaneously, and some babies learn to compensate adequately with the reduced mobility.[25,37] Only symptomatic cases that cause functional problems should be treated surgically. Treatment remains controversial, however, and there is no consensus regarding the indications, timing, or methods of surgical repair. Subjective outcomes show that in many cases, the division of a tongue-tie improves the mother's experience of breastfeeding.

Two systematic reviews have evaluated the effectiveness of division.[38,39] They identified five randomised controlled trials with heterogeneous and short-term outcome measures (Table 4[2,23,24,40,41]). The others were retrospective cohort studies and case series.

In their UK-based study of 201 babies, Hogan et al concluded that division is safe and improves feeding for both mother and baby.[2] In a US randomised, single-blinded controlled trial by Buryk et al,[23] the release of the tongue-tie reduced pain in the nipples and improved breastfeeding scores.

The National Institute for Health and Care Excellence (NICE) guidelines for the division of tongue-tie (2005) recommends early division (between one day and 12 weeks) for problems with breastfeeding or restricted mobility of the tongue. It can be done safely without anaesthesia, a recommendation that is endorsed by NICE.[36] General anaesthesia should be reserved for older infants or children, as the hazards of operation or reoperation are higher, ranging from 6% in infants (1-23 months) to 16% in children aged between 6 and 12 years.[36]

The current guidelines by the American Academy of Pediatric Dentistry recommend frenuloplasty for the management of problems with breastfeeding. For other functional problems such as speech, decisions should be made on a case-by-case basis.[10]

Terms that are used to refer to the division of tongue-tie are confusing, as they are interchangeable and inconsistent.[26] *Frenotomy* or *frenulotomy* refers to division of the lingual frenulum without sutures; *frenuloplasty* to division with the placement of sutures; and *frenectomy* or *frenulectomy* to excision of the lingual frenulum.

Frenotomy (frenulectomy) involves an incision of several millimetres into the lingual frenulum, which is a brief and usually bloodless procedure, as the frenulum in infants tends to be thin and relatively avascular. Haemostasis, if needed, is achieved by breastfeeding, which also lengthens the tongue and acts as an analgesic and antiseptic.[21] In infants less than 3 months of age, division is quick and simple, and can be done without anaesthesia in outpatients (Fig. 1).[8,42]

Complications are rare, but can include bleeding, infection, injury to the musculature of the tongue, and damage to the submandibular ducts and orifices.[42–44] The tie can recur in young infants after frenotomy, mostly as a result of excessive scarring.[40]

Table 3
Bristol Tongue Assessment Tool.

	Score		
	0	1	2
Tongue tip appearance	Heart-shaped	Slight cleft/notched	Rounded
Attachment of frenulum to lower ridge	Attached at top of gum ridge	Attached to inner aspect of gum	Attached to floor of mouth
Lift of tongue with mouth wide (crying)	Minimal tongue lift	Edges only mid-mouth	Full tongue-lift to mid-mouth
Protrusion of tongue	Tip stays behind gum	Tip over gum	Tip can extend over lip

Table 4
Randomised controlled trials to investigate the efficacy of division of tongue-tie in neonates. All studies showed level 2 evidence.

First author, year, country, and reference	No. of neonates	Indications	Study	Outcome
Emond 2013 UK[40]	107	Breastfeeding at 5 days; painful nipples	Age: 8-16 days Sex: not specified Sample: frenotomy n = 55; controls n = 52	No difference in effectiveness of breastfeeding (latch, audible swallowing, nipple type, comfort, hold score)
Berry 2012 UK[24]	60	Poor latch, painful nipples/ trauma, inefficient feeding	Age: 5-115 days; Sex: M:F – 2:1 ratio Sample: frenotomy n = 27 (3 excluded); controls n = 30	Immediate improvement in breastfeeding in frenotomy (21/27) cf control (14/30) ($p < 0.02$) No difference in nipple pain score
Buryk 2011 United States[23]	58	Breastfeeding problems, painful nipples	Age: 6 days Sex: M:38 F:20 Sample: frenotomy n = 30; controls n = 28	Reduction in nipple pain in frenotomy group cf control group ($p < 0.001$)
Dollberg 2006 Israel[41]	25	Painful nipples	Age: 1-21 days Sex: not specified Sample: frenotomy n = 14; controls n = 11	Improvement in pain score after frenotomy ($p = 0.001$) Significant improvement in latching ($p = 0.06$)
Hogan 2005 UK[2]	57	Poor latch, inefficient feeding, pain, mastitis, bottle-feeding problems	Age: frenotomy: median = 14 days; controls: median = 15 days Sex: study group: M:F – ratio 1:1; controls: M:F – ratio 3:1 Sample: frenotomy n = 28; controls n = 29	Frenotomy offered at 48 hours showed improvement in study group (27/28) cf control group (1/29) ($p < 0.001$)

Cf: compared with.

The role of an adviser on infant feeding

Advisers on infant feeding or lactation consultants have a pivotal role after lingual frenotomy, and studies show that the support they give improves the experience of breastfeeding and promotes positive outcomes.[45,46] Increasing the mother's awareness of the importance of positioning and maintaining a good latch is vital after the release of a tongue-tie, and will help the infant to adapt.

The future

Current evidence suggests that symptomatic ankyloglosssia will interfere with breastfeeding. Treatment is simple and effective without complications and there is a minimal cost to the public healthcare service. Knowledge of the condition by healthcare professionals and its potential effect on breastfeeding, however, seems to be limited, and seems to coincide with patchy provision of a service that enables prompt treatment.

In the first 30 days of life, infants are cared for by their parents, midwives, paediatricians, and healthcare visitors. We think that the provision of training and education would enable early identification of treatable symptomatic ankyloglossia. An agreed, universal, and simple questionnaire, which could be used to assess functional problems during the routine examination of neonates by midwives and health visitors, could easily be implemented and disseminated after training.

Increased awareness and a multidisciplinary approach will facilitate a more streamlined service. Encouragingly, despite a time of austerity, the NHS has responded to reports of national need and has facilitated the infrastructure for tongue-tie services across the UK (Table 5).[47]

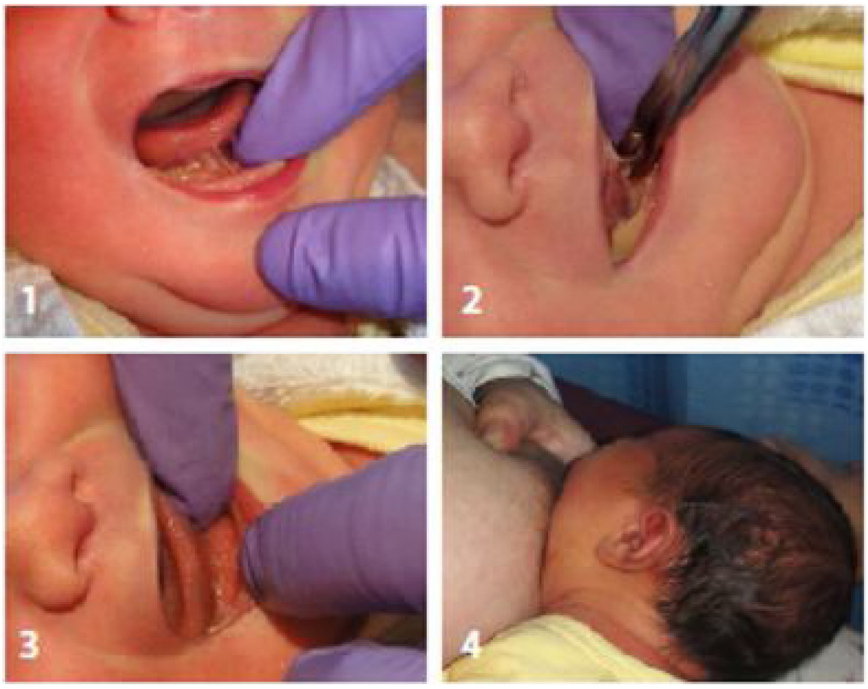

Fig. 1. Preoperative neonatal ankyloglossia (1); division with McIndoe scissors (2); free movement of the tongue postoperatively (3); and immediate breastfeeding with good suction from a fully functioning tongue (4) (photographs published with the parents' permission).

Table 5
Responses to a survey of the provision of tongue-tie services in the UK by the National Childbirth Trust (NCT), and current provision.[47]

	No. of Trusts with maternity units	No. of Trusts that responded	No. of Trusts with tongue-tie service
England	141	71	58
Scotland	14	5	3
Wales	7	5	5
Northern Ireland	5	5	5

Conflict of interest

We have no conflicts of interest.

Ethics statement/ confirmation of patients' permission

Ethics approval N/A. Parental consent was gained for the images used.

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