Ankyloglossia: A Review of Current Status
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Citation

Abstract
Ankyloglossia, ankyloglossia inferior or tongue tie is an uncommon but not rare congenital anomaly. It is characterized by a thick short lingual frenulum which restricts the mobility of the tongue. This can lead to a range of problems such as difficulties in breast feeding in infancy, speech articulation defects and various psychosocial problems in childhood, adolescent and in adults. However there appears to be considerable diversity in the criteria for diagnosis and management of ankyloglossia. A review of the current opinion on ankyloglossia is presented.

INTRODUCTION
Ankyloglossia is a congenital anomaly characterized by presence of a hypertrophic lingual frenulum which is short and attached to the very tip of the tongue, limiting its normal movements. Clinically the patient cannot protrude the tongue past the incisal edge of the gingiva and the tongue becomes heart shaped when attempted to protrude. The restricted movements of the tongue can result in problems with breast feeding, lactation, speech disorders and other oral motor disorders like problems with swallowing and licking. The clinical significance of this anomaly and symptoms produced and the best method of management have been the subject of debate for some time. A review of the relation between the above problems and ankyloglossia is presented.

REVIEW
Tongue is an accessory organ of importance in mastication, deglutition and speech; many authors describe its stimulatory influence on the development of the dental arches. At birth, the tongue unconfined by the teeth, extends outward between the maxillary and mandibular occlusal gum pads. The “infantile swallow” with jaws parted and tongue placed between the jaws is replaced by the adult swallow at the age of two and half years of age. At the start of the normal adult swallow, the lips are closed, the teeth are brought into full occlusion, and the tip is raised and placed against the anterior portion of the palate. The respiratory opening, the nasal cavity, and the anterior portion of the mouth are all sealed off as the tongue, in a sweeping and undulating motion, sweeps backward the mass of chewed food. The tongue is always short at birth and the tip of the tongue is yet incompletely developed. As the infant grows, the tongue becomes longer and thinner towards the tip.

Tuerk and Lubit proposed two dental deformities as a consequence of ankyloglossia. Open bite deformity and mandibular prognathism. The inability to raise the tongue to the roof of the palate encourages the continuation of the infantile swallow, prevents development of the adult swallow and leads to an open bite deformity. The lack of a free upward and backward movement of the tongue which may result in an exaggerated anterior thrusting of the tongue against the anterior body of the mandible, and produce mandibular prognathism. Horton et al reported that the prominent lower frenulum may lead to repeated lower denture plates dislodgement when the tongue is elevated. The above possibility was also noted by other authors.

INCIDENCE AND SEX RATIO
Incidence of ankyloglossia reported in the literature varies widely ranging from 0.02% to 4.8%. The variation may be attributed partly to the lack of a uniform definition and objective grading system for tongue tie. It occurs most commonly in males with a male to female ratio of 3 to 1, and shows no racial predilection. Ankyloglossia occurs most frequently as an isolated anatomic variation. Increased incidence of ankyloglossia is noted in infants with a history of maternal cocaine abuse, in association with various congenital syndromes, including Opitz syndrome, orodigitofacial syndrome and X-linked cleft palate.
EFFECT ON SPEECH

The effect of ankyloglossia on speech has not been clearly defined. Speech problems historically have been viewed as the hallmark manifestations of symptomatic tongue-tie, particularly among the lay public. Contrary to public opinion, tongue tie have comparatively slight effects on speech. Anecdotal evidence indicates that some children with ankyloglossia develop normal speech, and compensate for limited tongue tip mobility without surgical repair or need for speech therapy.\textsuperscript{13,17} In some cases tongue-tie contributes to articulation errors. Speech sounds that may be affected by impaired tongue-tip mobility include lingual sounds and sibilants such as T, D, Z, S, TH, N and L.\textsuperscript{3,13,14} The compensatory techniques used by children with ankyloglossia typically include restricted mouth opening while speaking, and alternate tongue placement for sounds requiring tongue-tip elevation.\textsuperscript{15} The compensation may be affected by factors such as dentures, missing incisors, tongue size, sensory and motor function of the tongue, as well as the degree of ankyloglossia.\textsuperscript{9} While not preventing the acquisition of speech at a normal age, the shortened frenulum may sometimes cause minor difficulties, such as minor speech defects. The production of certain consonants is effected by a tongue which is not mobile enough to achieve tongue tip or dorsal-alveolar ridge contact.\textsuperscript{16}

Fletcher and Meldrum measured the anterior, inferior segment of the tongue and the relation of such measurements to the physiological performance of the tongue. Forty 11 to 12 year old children with no speech defects were selected and placed in two groups on the basis of anatomical sublingual measurements: the limited lingual freedom group and greater lingual freedom group. A modified Boley gauge was used for measurement. Each group was administered the 176-item Templin-Darley articulation test. The authors found a highly significant increase in the articulation errors in the limited freedom as compared with the greater lingual freedom group. They suggested that significant interaction exists between lingual mobility and speech proficiency.\textsuperscript{17}

The severity of the articulation problems in affected individuals vary from being evident at the single word level or mild enough so as to be noticed only in connected speech. It is important to remember that ankyloglossia is not a cause of speech delay. Such children are expected to acquire speech and language at a normal rate, although some may experience articulation difficulties for certain speech sounds. In a child with delayed speech with tongue tie the potential causes of speech delay should be carefully sought and the patient should be directed for other evaluations.\textsuperscript{15,17}

MECHANICAL AND SOCIAL EFFECTS

Tongue tie have potential mechanical and social consequences which include: difficulty with intraoral toilet, licking the lips and sweeping the teeth free of food debris, difficulty in wearing dentures later in life due to poor fit, decreased facility in playing a wind instrument, difficulty in licking an ice-cream and difficulty with “French-kissing”.\textsuperscript{5,13,18} Further these symptoms may be accompanied by a sense of social embarrassment due to teasing and ridicule from peers.\textsuperscript{19,20} The mechanical and social concerns may not manifest until later in childhood as younger children indeed may have symptoms but may not recognize or able to report them. Moreover owing to a sense of embarrassment regarding their condition patients may refrain from expressing their concerns unless questioned directly.\textsuperscript{15}

EFFECT ON BREAST FEEDING

For more than 4200 species of mammals, breastfeeding is a natural process ensuring survival of species.\textsuperscript{21} Breast feeding should be initiated within an hour of birth. The first milk called colostrum is the most suitable food for the newborn. Breast milk is a complex fluid containing numerous anti-infective agents and hormones. The various advantages of breast milk include: its safe and hygienic and available at correct temperature, it fully meets the nutritional requirements of the infant in first few months of life, it contains antimicrobial factors such as macrophages, lymphocytes, secretary IgA, anti-streptococcal factor, lysozyme and lactoferrin which provides protection against diarrhea and respiratory infections. Further it promotes bonding between the mother and the infant.\textsuperscript{22} Colostrum prevents renal overload as the newborn baby is adjusting its fluid balance. Its potential long term health benefit in children includes reduced blood pressure, cholesterol concentrations and obesity levels. Further breast milk reduces morbidity and mortality rates.\textsuperscript{23}

Ankyloglossia has been linked to breast feeding difficulties. The swallowing mechanism of the new born and infant is different from the adult or older children. The infant must latch on to the mother’s areola with his/her tongue. Suckling begins with the forward movement of the jaw and tongue. The tongue helps to make a better seal, with minimal active action. The anterior edge of the tongue thins, cupping upwards to begin a peristaltic ripple back towards the throat. The lower jaw at this juncture squeezes milk from the ductules and finally, the posterior part of the tongue
depresses to allow milk to collect in the oropharynx before swallowing.\textsuperscript{2,17,21,24} Thus restrictions of the tongue movements due to tongue-tie may be quite extreme to interfere with suckling and swallowing.

Most lactation consultants and some physicians have found that tongue tie can make breast feeding difficult, causing sore nipples, poor infant weight gain, low milk supply with early weaning and maternal fatigue and frustration.\textsuperscript{25-27} Ankyloglossia seems to have more deleterious effect on the breast feeding than on bottle feeding because of the relative difference in tongue movements with breast and bottle feeding.\textsuperscript{28-31} Multiparous mothers who have breast-fed their unaffected infants successfully in the past note an obvious difference when nursing a subsequent newborn with tongue-tie. Moreover, mothers of affected infants frequently report a marked improvement in breast-feeding after tongue tie release.\textsuperscript{15} There is general agreement that infants with ankyloglossia do not have trouble with bottle feeding, nor with handling solid foods when they are introduced. Bottle feeding however should not be proposed as a solution for ankyloglossia related problems in a mother who otherwise desires to breast feed.\textsuperscript{15,28,30}

Miranda and Milroy in a prospective study of 62 neonates of ankyloglossia and breastfeeding difficulties and reported that at two weeks post-frenulotomy, there were significant improvements in weight and breastfeeding, including number of sessions per twenty four hours and bottle top-ups per twenty four hours.\textsuperscript{32}

Khoo et al conducted a prospective cohort study to test the hypothesis that in mother-infant pairs experiencing breastfeeding difficulties, frenulotomy for tongue tie may improve breast feeding. Sixty two infants under 90 days underwent frenulotomy. At presentation fifty two mothers reported nipple pain and thirty two nipple trauma. Those having difficulty in breast feeding due to nipple pain showed a significant long term benefit from frenulotomy; pre-frenulotomy nipple pain was associated with an increased likelihood of breastfeeding at three months.\textsuperscript{33}

Dollberg et al in a study of 25 mothers of healthy infants with ankyloglossia reported that a significant decrease in nipple pain score occurs after frenotomy.\textsuperscript{34} Similar results were also reported by Wallace and Clarke\textsuperscript{35}, Hogan et al\textsuperscript{36} the authors further stated that tongue tie can affect feeding and that division of tongue tie is safe, successful and easy procedure and infants with a significant tongue tie that is interfering with breast feeding show improvement in breast feeding after frenotomy.

**DIAGNOSIS**

The diagnosis of ankyloglossia may be difficult, especially in infants, since the condition is not always apparent on observation of the undersurface of the tongue. In infants a passive elevation of the tip of the tongue blade may demonstrate the short frenulum which restricts the mobility of the tip.\textsuperscript{(Fig 1)}

**Figure 1**

Fig 1: Ankyloglossia demonstrating the short frenulum

Palpation of the tight genioglossus muscle on the undersurface of the tongue will help to confirm the diagnosis.\textsuperscript{9} When the child is old enough to cooperate, moving the tongue through its maximum range will reveal tip restriction. The tip of the tongue cannot be protruded beyond the lower incisor teeth in true tongue tie because of the short frenulum linguae. Attempt to protrude the tongue produces notching of its tip or heart shape of tongue, due to tethering by the frenulum.\textsuperscript{13,15} Protrusion is limited and in some cases may fail to extend past the lower lip. The tongue may appear to roll or curl with attempted protrusion as the mid portion of the tongue moves forward while the tip itself is drawn inferiorly by the frenulum with little forward extension.\textsuperscript{(Fig 2)} There is difficulty in lifting or elevating the tip of the tongue toward the upper dentition and upper lip. In most cases patient can raise the tip to some degree, but it is noted that while the tip may point straight up, the dorsum of the tongue remains flat and there may often be a dimpling in the dorsal area which corresponds to the insertion of the restraining genioglossus muscle.\textsuperscript{15}
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Figure 2
Fig 2: The tongue may appear to roll or curl with attempted protrusion.

A space or diastema between the lower teeth is occasionally present, due to the repeated thrusting action of the frenulum through the teeth with attempted tongue protrusion. Various schemes have been proposed to objectively verify the presence of ankyloglossia. Assessment tool of lingual frenulum function was devised by Hazelbaker to evaluate the tongue movements, including lateralization, lift, extension, spread of anterior tongue, cupping, peristalsis and snap-back. Each tongue movement is graded on 0 to 2 scales. Poorly scoring patients are advised frenotomy. Kotlow presented a simple classification in which free tongue is measured. The free tongue length is the distance in mm from tongue tip to the insertion of the frenulum. The tongue-tie can be classified into four classes: mild, moderate, severe and complete depending upon the measured free tongue length of 12-16 mm, 8-11 mm, 3-7 mm and < 3 mm respectively. This classification, however, failed to provide any documentation regarding the correlation of ankyloglossia class to the clinical symptoms. Williams and Wardon proposed a method to measure the lingual function instead of physical length. The patient is asked to touch the tip of his/her tongue to the upper teeth, and then open the mouth as widely as possible while maintain this contact. In this position the distance between the upper and lower central teeth i.e the inter-incisal distance is recorded in millimeters. Lalakea and Messner measured lingual mobility in children for protrusion and elevation and reported that protrusion and elevation values are typically in the range of 15 mm or less in children with tongue tie and 20 to 25 mm or greater in normal children.

Ruffoli et al in a study of 200 children referred for evaluation of tongue tie measured the length of the frenulum and the inter-incisal distance in maximal opening of the mouth and with the tip of the tongue touching the palatal papilla. The authors reported that children with a frenulum length of more than 2 cm and an inter-incisal distance of more than 2.3 cm were normal. It should be remembered that, despite all the methods mentioned, at present there is no way to exactly predict on the basis on examination findings which children are likely to develop speech or mechanical symptoms due to ankyloglossia. However mobility measurements are a valuable tool in documentation of change particularly preoperatively and postoperatively.

TREATMENT

History and physical examination are sufficient to confirm the diagnosis of ankyloglossia. The patient’s ability to protrude the tongue past the lower lip has been used by some in the past as a quick rule of thumb to predict which patients will not require surgical intervention. Patients with ankyloglossia with obvious or definite indications for surgery include infants with associated breast feeding difficulties, young children with characteristic articulation problems, and older children and adults adversely affected by the mechanical and social manifestations of tongue tie. Tuerk and Libilit stated that ankyloglossia should be corrected, when associated with an open bite or mandibular prognathia, prior to the corrective orthodontic care. Further even a well compensated ankyloglossia may require surgery prior to the construction and fitting of dentures in an edentulous mouth.

The optimal timing for the surgery has not been determined. Some authors advocates surgical intervention prior to the development of speech difficulties, while others suggest withholding surgery until age four and offering surgery only to those who manifest speech problems. Lalakea and Messner suggest that given the minor nature of the surgery and significant potential for speech difficulties and later social and mechanical problems, it may be appropriate to consider surgery for those children with significant ankyloglossia at any age, including infants and toddlers who have yet to demonstrate overt symptoms. Some authors though feel that surgery be deferred until the age of four years.

SURGICAL PROCEDURES

Several surgical procedures have been used to achieve release of ankyloglossia. The midwives in past, used their
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fingernail for ripping of the frenulum. The procedures frenotomy or simple release of the frenulum and frenuloplasty (release with plastic repair) are the two most commonly used surgical procedures in the treatment of ankyloglossia.\textsuperscript{15}

Frenotomy is most appropriate for the treatment of ankyloglossia in infants. The procedure is rapid and easy to accomplish and can be performed at the bedside in the newborn nursery, or in the office with local or no anesthesia. The parent sits opposite the doctor, with knees in contact and the infant lies supine with his/her head towards the doctor. The tongue is lifted superiorly with the doctor’s gloved middle and index fingers. The doctor uses the other hand to divide the tongue tie with a pair of blunt-tipped scissors.\textsuperscript{15}

Griffiths conducted a study of 215 infants younger than 3 months (mean 0-19 days) having major problems with breast feeding despite professional support. The author reported that prior to division 88% had difficulty in latching, 77% mothers experienced nipple trauma and 72% had a continuous feeding cycle. Within 24 hours of division 80% were feeding better.\textsuperscript{15}

Frenuloplasty is preferred for older than 1-2 years of age. In young children, the procedure is performed under general anesthesia while in older children and adults it can be done under local anesthesia. The frenulum is released in a manner identical to frenotomy. A limited division of the genioglossus muscle may however be required for complete release.\textsuperscript{44} Frenotomy under no or local anesthesia is safe and cost effective, but one third of children need re-operation.\textsuperscript{15}

Heller et al in a study of 16 patients more than three years of age with persistent articulation problems related to ankyloglossia underwent surgical correction. A 4-flap Z-frenuloplasty was performed in 11 cases and traditional horizontal to vertical frenuloplasty in five cases. The authors reported that 4-flap Z-frenuloplasty was superior to vertical frenuloplasty with respect to tongue lengthening, protrusion and articulation improvement for patients with symptomatic ankyloglossia.\textsuperscript{15}

The patients old enough to comply are asked to perform tongue exercises in order to enhance mobility, assist in retaining the tongue musculature and reduced potential of scarring.

**COMPlications**

An extensive division of the frenulum may injure the deep vessels of the tongue resulting in hemorrhage. Postoperative scar contracture may result in more deforming ankyloglossia. There may be ulceration and infection of the floor of the mouth and occasionally hyper-mobility of the tongue and secondary attacks of asphyxia from glossoptosis.\textsuperscript{13,46} Walsh and Kelly reported a case of a seven year old boy who developed evidence of upper airway collapse which resolved spontaneously within an hour. The authors stated that a tight frenulum also holds the tongue anteriorly and after surgical release the genioglossus muscle may not be able to generate sufficient force to prevent airway collapse.\textsuperscript{47}

Recent advances for correction of ankyloglossia

Monopolar diathermy has been used to for frenotomy under local anesthesia for release of tongue tie.\textsuperscript{48} Aras et al compared diode laser and erbium:yttrium-aluminium-garnet (Er:YAG) lasers for the tolerance of postsurgical discomfort experienced by patients operated for tongue tie. They reported that there was no difference between the two groups regarding the postsurgical discomfort except in the first three hours in which Er:YAG group patients had more pain. However majority of the patients (six) could be operated without local anesthesia in Er:YAG group, all patients could not be operated on without local anesthetic in the diode group, suggesting that Er:YAG laser is more advantageous than diode laser in minor soft tissue surgery.\textsuperscript{49} Verco used Explor Ar plasma cutting electrode (APCE) with ConMed Argon beam coagulator in tongue tie and reported that it is helpful for bloodless approach to the treatment of tongue tie with little or no pain.\textsuperscript{50}

**References**

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