

Provided for non-commercial research and education use.
Not for reproduction, distribution or commercial use.



This article appeared in a journal published by Elsevier. The attached copy is furnished to the author for internal non-commercial research and education use, including for instruction at the authors institution and sharing with colleagues.

Other uses, including reproduction and distribution, or selling or licensing copies, or posting to personal, institutional or third party websites are prohibited.

In most cases authors are permitted to post their version of the article (e.g. in Word or Tex form) to their personal website or institutional repository. Authors requiring further information regarding Elsevier's archiving and manuscript policies are encouraged to visit:

<http://www.elsevier.com/copyright>



Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Case report

Posterior ankyloglossia: A case report

Michael W. Chu^{a,*}, David C. Bloom^b^a Department of Otolaryngology – Head & Neck Surgery, Eastern Virginia Medical School, 600 Gresham Drive, Suite 1100, Norfolk, VA 23507, USA^b Department of Otolaryngology – Head & Neck Surgery, United States Naval Hospital, Okinawa, Japan

ARTICLE INFO

Article history:

Received 6 October 2008

Received in revised form 10 February 2009

Accepted 12 February 2009

Available online 20 March 2009

Keywords:

Posterior ankyloglossia

Tongue-tie

Frenulectomy

Frenuloplasty

ABSTRACT

Ankyloglossia, or tongue-tie, refers to an abnormally short lingual frenulum. Ankyloglossia is a recognized but poorly defined condition and has been reported to cause feeding difficulties, dysarthria, dyspnea, and social or mechanical problems. In infants, the most concerning symptoms are feeding difficulties and inability to breastfeed. While a recent trend toward breastfeeding has brought frenulectomy back into favor, the literature regarding treatment remains inconclusive. We report a case of posterior ankyloglossia with anterior mucosal hooding and a simple, safe, and effective way to treat it to improve breastfeeding.

© 2009 Elsevier Ireland Ltd. All rights reserved.

1. Introduction

Ankyloglossia, or tongue-tie, refers to an abnormally short lingual frenulum. Ankyloglossia is derived from the Greek words 'ankylo' meaning stiff and 'glossa' meaning tongue. However, there remains controversy concerning the precise definition of ankyloglossia, its causal relationship to infant symptoms, and its management.

Ankyloglossia 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 [1]. The Academy of Breastfeeding Medicine Protocol defines ankyloglossia 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 into the gingival ridge' [2]. The reported prevalence of ankyloglossia varies from 0.02 to 4.8% [2], but only causes feeding difficulties in 44% [3]. The Hazelbaker Assessment Tool for Lingual Frenulum Function [4] (HATLFF) was developed to provide a quantitative assessment of ankyloglossia and has been proven to be highly reliable. It includes five appearance items, such as length, attachment site, and elasticity, as well as seven functional items, such as extension, spread, cupping, and peristalsis of the tongue [2,4,5].

Ankyloglossia has been reported to cause feeding difficulties, dyspnea from forward dislocation of the epiglottis and larynx;

speech articulation problems involving lingual alveolar sounds /l/ and interdental sounds /th/; and social and mechanical problems (inability to lick lips, maintain oral hygiene, play wind instruments, enjoy ice cream cones, blow bubbles, and French kiss) [1,6]. In infants, the most concerning symptoms are breastfeeding difficulties related to ineffective latching, decreased ability to create a seal, poor weight gain, and maternal nipple pain.

The management of ankyloglossia varies among different health care providers. Pediatricians, pediatric surgeons, otolaryngologists, dentists, lactation specialists, and speech language pathologists [6] are all involved in the care of tongue-tie, but there is little consensus regarding the significance in symptomatology, its causal relationship to dysfunction, and its management.

In the early 1900s, tongue-tie was believed to impact breastfeeding and was routinely divided. As formula milk gained popularity, tongue-tie release fell out of favor as infants could bottle feed as an alternative [7]. Recent literature disputes that tongue-tie has any effect in infant feeding or speech [6], and instead recommends feeding specialist consultations, bottle-feeding, and conservative, non-surgical management options. The benefits of breastfeeding are well established, and the new trend to encourage breastfeeding coupled with increased awareness of ankyloglossia has again brought tongue-tie release back into discussion [3,6,8]. The literature remains inconclusive with only anecdotal reports or limited prospective, controlled studies suggesting that tongue-tie can influence infant breastfeeding as well as maternal discomfort [3,7,8].

The history of ankyloglossia correction dates back to the New Testament: "one... had an impediment in his speech... the string was of his tongue was loosed, and he spake plain." (Mark 7:32). In

* Corresponding author. Tel.: +1 757 388 6200; fax: +1 757 388 6201.

E-mail address: chumw@evms.edu (M.W. Chu).

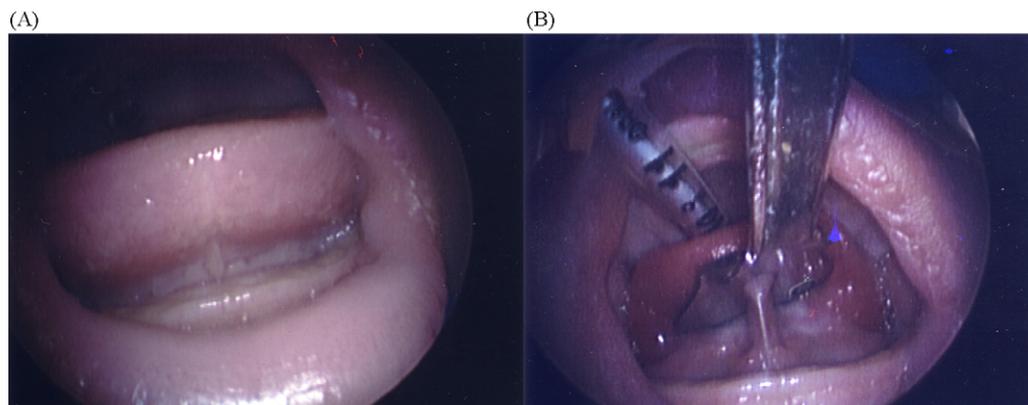


Fig. 1. Four-week-old infant with a short frenulum posterior to the anterior “mucosal curtain.” (A) Initial assessment does not show a remarkable frenulum or tongue-tie. (B) After retraction posteriorly of ventral tongue mucosal with a groove director, the frenulum is noted to be a short, thick, fibrous cord posterior to the ventral tongue mucosa, causing significantly tongue-tie which was otherwise obscured by the “mucosal curtain.”

the 18th century midwives would use their fingernails to divide the frenulum, and there has since been a variety of descriptions, including simple division, frenulotomies, and four-flap Z-frenuloplasty [1]. Tongue-tie release can be performed safely, quickly, and without anesthesia in infants less than 3 months of age [3,5,8] and is a possible treatment for feeding difficulties. We present a new condition described as a posterior ankyloglossia, due to the location of the frenulum posterior to the anterior mucosal covering of the ventral tongue and floor of mouth. A frenulum that is posterior to the mucosal covering can be hidden and lead to difficulty in diagnosing a condition that can otherwise be safely and effectively treated.

2. Case report

We report a rare entity we describe as a posterior ankyloglossia with anterior mucosal hooding. A 4-week-old infant male was referred by a lactation specialist for difficulty with feeding as well as pain with breastfeeding. Initial physical exam did not reveal an obvious ankyloglossia or prominent frenulum. But on palpation and retraction with a groove director, the frenulum was noted to be a fibrous cord posterior in relationship to the anterior mucosa of the tongue (Fig. 1). The frenulum was thus obscured by what we describe as a “mucosal curtain” from the anterior tongue to the floor of mouth that would not have been noticed without retraction. The short frenulum was thought to cause tongue-tie that was interfering with parental desire to breastfeed the patient. Given the symptomatology and physical exam findings, the patient was taken to the operating room for a horizontal-to-vertical frenuloplasty as previously described [9].

The patient underwent general anesthesia and the “mucosal curtain” was retracted with a groove director, which revealed a significant tongue-tie and short frenulum.

The frenulum was a short fibrous cord posterior to the anterior mucosa, restricting full range of motion of the tongue. Local anesthetic was injected in the frenulum and lateral mucosa. A straight hemostat was then clamped parallel to the tongue at the ventral attachment of the frenulum. This is our standard practice for bedside frenulotomies and provides excellent hemostasis after incision. The clamp was then released and straight scissors were used to release the frenulum (Fig. 2). The tongue was further retracted superiorly and on palpation, there was persistent tethering of the tongue by the fibrous cord of the frenulum. The frenulum was clamped again and incised. Dissection was carried posteriorly with care to avoid a fistula in the floor of mouth inferiorly and bleeding in the vascular tongue musculature superiorly. A combination of blunt and sharp dissection freely

mobilized the entire anterior tongue (Fig. 3A), and on palpation there was no further evidence of a fibrous cord or tethering.

The diamond-shaped mucosal defect was closed with 4–0 chromic sutures in a horizontal-to-vertical frenuloplasty (Fig. 3B). The most distant lateral apexes were first sutured closed with a simple interrupted stitch, which caused an immediate advancement and anterior protrusion of the tongue beyond the incisors. Three additional sutures were placed to close the mucosal defect. There was excellent hemostasis throughout the case and negligible blood loss. The patient tolerated the procedure well and there were no complications. On follow-up there was decreased maternal nipple pain and the patient was able to successfully breastfeed.

3. Discussion

Ankyloglossia is a congenital abnormality of the lingual frenulum. The exact pathophysiology of tongue-tie is unknown. The mucosa covering the anterior two-thirds of the mobile tongue is derived from the first pharyngeal arch, and deviation of normal development is the most likely cause of abnormal frenulum length and attachment. There are several syndromes associated with the physical finding of ankyloglossia, including Ehlers-Danlos syndrome, Beckwith-Wiedemann syndrome, Simosa syndrome, X-linked cleft palate, orofaciogigital syndrome, and several others [6,10]. Ehlers-Danlos syndrome (EDS) may provide insight in understanding ankyloglossia as it includes a group of more than

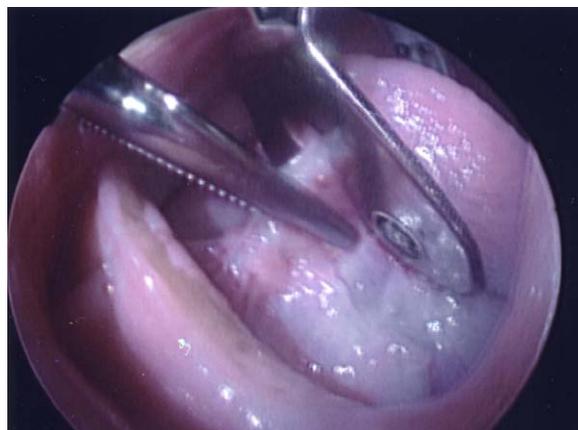


Fig. 2. The frenulum is clamped inferior to groove director, parallel to the attachment to ventral tongue, for 5 s for hemostasis. Clamping and crushing the frenulum before incision is an effective method for achieving hemostasis and can also be performed at the bedside.

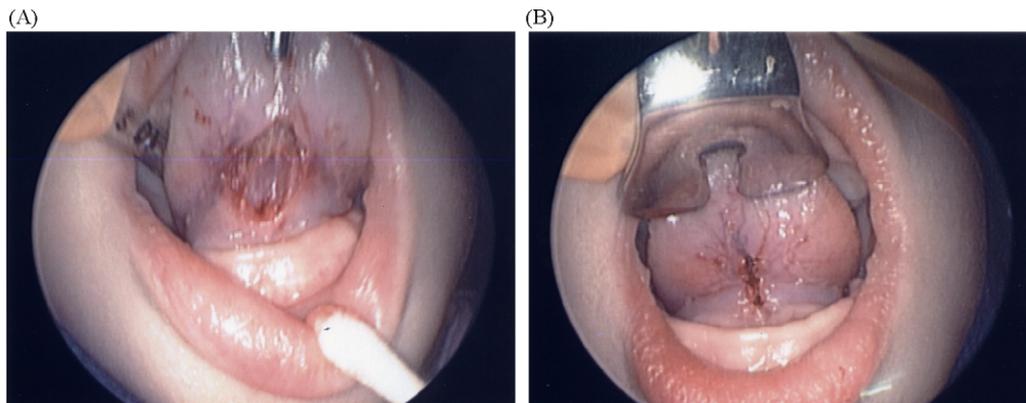


Fig. 3. (A) The frenulum is released with sharp dissection resulting in a diamond-shaped mucosal defect. Care is taken to prevent excessive blood loss by avoiding the vascular, intrinsic muscles of the tongue and to prevent potential fistula by avoiding dissection into the floor of mouth. (B) Horizontal-to-vertical frenuloplasty is performed with release of tension and attachment to the frenulum. There was immediate anterior protrusion and increased mobility of the tongue.

ten inheritable connective tissue disorders. EDS is characterized by skin hyperextensibility and joint hypermobility due to collagen and extracellular matrix defects. Ankyloglossia has been shown to have a strong family history and the frenulum is a connective tissue structure, thus it is likely related to abnormal development of collagen and connective tissue in the anterior tongue.

Posterior frenula may often be missed due to the position of the ventral tongue mucosa anterior to the fibrous cord, creating a “mucosal curtain.” But a thorough physical exam, including mucosa retraction posteriorly, will reveal any frenula obscured by the mucosal draping. Frenuloplasty is safe and effective method that can also treat posterior ankyloglossia and promote the benefits of breastfeeding.

4. Conclusion

Ankyloglossia is a recognized but vaguely defined condition. There continues to be debate concerning the pathophysiology, diagnosis, and management of tongue-tie. Posterior frenula with anterior mucosal hooding should also be considered in the differential diagnosis in patients without otherwise obvious ankyloglossia, and as a possible cause of feeding difficulties. Frenuloplasty is a safe, quick, and effective treatment that can provide immediate symptom relief, promote breastfeeding, and enhance infant-mother bonding experience.

Disclosure

None.

Disclaimer

The views expressed in this article are those of the author(s) and do not necessarily reflect the official policy or position of the Department of the Navy, Department of Defense, or the United

States Government. Dr. Bloom is a military service member. This work was prepared as part of his official duties. Title 17 U.S.C. 105 provides that “Copyright protection under this title is not available for any work of the United States Government.” Title 17 U.S.C. 101 defines a United States Government work as a work prepared by a military service member or employee of the United States Government as part of that person’s official duties.

Acknowledgements

The authors wish to thank both Jeannette Carter for her assistance in caring for children with ankyloglossia at Naval Medical Center Portsmouth and Dr. Elizabeth Coryllos for her expertise in the area of ankyloglossia and assistance with this case.

References

- [1] J. Heller, J. Gabbay, C. O'Hara, et al., Improved ankyloglossia correction with four-flap Z-frenuloplasty, *Ann. Plast. Surg.* 54 (June (6)) (2005) 623–628.
- [2] L.H. Amir, J.P. James, S.M. Donath, Reliability of the Hazelbaker assessment tool for lingual frenulum function, *Int. Breastfeed J.* 1 (March (1)) (2006) 3–9.
- [3] M. Hogan, C. Westcott, M. Griffiths, Randomized, controlled trial of division of tongue-tie in infants with feeding problems, *J. Paediatr. Child Health* 41 (May (5)) (2005) 246–250.
- [4] A.K. Hazelbaker, *The Assessment Tool for Lingual Frenulum Function (ATLFF) Use in a Lactation Consultant Private Practice*, Pacific Oaks College, Los Angeles, 1993.
- [5] L.H. Amir, J.P. James, J. Beatty, Review of tongue-tie release at a tertiary maternity hospital, *J. Paediatr. Child Health* 41 (5) (2005) 243–245, May.
- [6] A. Kummer, Ankyloglossia: To clip or not to clip? That's the question, *The ASHA Leader* 10 (December (17)) (2005) 6–7, 30.
- [7] H. Wallace, S. Clarke, Tongue tie division in infants with breast feeding difficulties, *Int. J. Pediatr. Otorhinolaryngol.* 70 (July (7)) (2006) 1257–1261.
- [8] J.L. Ballard, C.E. Auer, J.C. Khoury, Ankyloglossia: assessment, incidence, and effect of frenuloplasty on the breastfeeding dyad, *Pediatrics* 110 (November (5)) (2002) 63–69.
- [9] V. Velanovich, The transverse-vertical frenuloplasty for ankyloglossia, *Mil. Med.* 159 (November (11)) (1994) 714–715.
- [10] C. De Felice, P. Toti, G. Di Maggio, et al., Absence of the inferior labial and lingual frenula in Ehlers-Danlos syndrome, *Lancet* 357 (May (9267)) (2001) 1500–1502.