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# Ankyloglossia (tongue-tie) in infants and children

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### INTRODUCTION

Ankyloglossia, or tongue-tie, is a condition of limited tongue mobility caused by a restrictive picture 1A-B) [1]. The definition of ankyloglossia is not standardized, and there is wide variation of opinion regarding its clinical significance and optimal management [2,3].

Ankyloglossia will be discussed here. Other congenital anomalies of the tongue are discussed separately. (See "Congenital anomalies of the jaw, mouth, oral cavity, and pharynx".)

### **ANATOMY**

The lingual frenulum is frequently referred to as a "cord" or "submucosal band" of connective tissue. In fact, the lingual frenulum is a dynamic structure formed by a midline fold in a layer of fascia that inserts around the inner arc of the mandible, forming a diaphragm-like structure across the floor of mouth. This fascia is located immediately beneath the oral mucosa, fusing centrally with the connective tissue on the tongue's ventral surface. The sublingual glands and submandibular ducts are enveloped by the fascial layer, and anterior genioglossus fibers are suspended beneath it [4]. Thus, the configuration and contents of the lingual frenulum vary depending on force applied to the tongue. There is considerable individual variation in the appearance of the lingual frenulum under tension. It may insert on the tip of the mobile tongue or along its undersurface. In some healthy infants, no frenulum is seen connecting the floor of mouth and mobile tongue. In such infants, portions of the genioglossus can be drawn up into the fold, creating a "posterior" tongue-tie when the tongue is elevated.

Branches of the lingual nerve are located superficially on the ventral surface of the tongue, immediately beneath the fascia, making them vulnerable to injury during frenotomy procedures [5].

### **EPIDEMIOLOGY**

The reported prevalence of ankyloglossia varies from <1 to 10 percent, depending upon the study population and criteria used to define ankyloglossia [6-12]. A uniform definition and objective grading system for tongue-tie are lacking. Some grading systems, such as the Coryllos classification, focus on the type of frenulum but do not address functionality. Other systems, such as the Hazelbaker Assessment Tool for Lingual Frenulum Function and the Bristol Tongue Assessment Tool, attempt to include functionality and ankyloglossia scoring ( figure 1 and figure 2) [1].

In most series, the frequency of tongue-tie is higher among boys, with a male-to-female ratio of 1.5:1 to 2.6:1 [6]. While most cases of ankyloglossia are sporadic, mutations in the T box transcription factor TBX22 may lead to heritable (X-linked) ankyloglossia with or without cleft lip, cleft palate, or hypodontia [13].

### CLINICAL FEATURES

**Ankyloglossia** — Clinical features of ankyloglossia may include [6,9,12,14]:

- Abnormally short frenulum, inserting at or near the tip of the tongue ( picture 1A-B)
- Difficulty lifting the tongue to the upper dental alveolus (
- Inability to protrude the tongue more than 1 to 2 mm past the lower central incisors ( picture 3)
- Impaired side-to-side movement of the tongue
- Notched or heart shape of the tongue when it is protruded (
- For infants too young to voluntarily protrude the tongue, a frenulum that prevents placement of the examiner's fingers between the underside of the tongue and mandibular alveolus is considered abnormally restrictive

The natural history of untreated ankyloglossia is unknown [14]. Some postulate that progressive stretching and use of the frenulum leads to spontaneous elongation, but this has not been substantiated in prospective studies [14].

**Posterior ankyloglossia** — The definition of "posterior" ankyloglossia is not standardized. Some experts use the term to mean a frenulum that inserts into the posterior portion of the tongue, whereas others think of it as a submucosal tethering of the tongue. Some experts question its existence as a real anatomic entity and advocate for abandoning the term "posterior ankyloglossia" [1].

**Upper lip tie** — There is controversy regarding the identification, classification, and significance of the superior labial frenulum in newborns as well as when the frenulum constitutes a "lip-tie." Existing rating systems have poor reproducibility, even when used by experienced clinicians [15]. All newborns have a labial frenula, with most attached at the gingival margins [16]. Rarely, infants may not effectively latch to the maternal breast due to inadequate eversion of the upper lip. Division of the upper labial frenulum has been advocated in such cases. The efficacy and appropriate indications for upper labial frenotomy are unknown [17]. (See 'Frenotomy' below.)

## **POTENTIAL SEQUELAE**

When symptoms related to ankyloglossia develop, they most commonly present in one of the following ways [14,18]:

- Newborn or young infant with breastfeeding difficulty (see <u>'Breastfeeding problems'</u> below)
- Toddler or young child with articulation problems (see 'Articulation issues' below)
- Older child or adolescent with mechanical difficulties and/or social embarrassment due to restricted tongue movement (eg, difficulty licking lips, difficulty kissing) (see 'Mechanical and social problems' below)

The extent to which tongue-tie results in clinically significant symptoms is controversial [14]. There is little high-quality evidence regarding this issue, with published information consisting largely of case series and observational studies [6,19]. Despite this, the rates of diagnosis of ankyloglossia and referral for surgical treatment have nearly doubled since the early 2000s, with wide regional variation [20].

**Breastfeeding problems** — The majority of infants with tongue-tie are able to breastfeed figure 4) [8]. However, breastfeeding problems (eg, poor latch, maternal without difficulty ( nipple pain) are reported more frequently among infants with ankyloglossia than without ankyloglossia (25 versus 3 percent in one series) [8,12]. Poor latch with resultant low milk transfer can lead to failure to thrive in the exclusively breastfed infant [12,21]. Studies have not found any effect of ankyloglossia on bottle feeding, most likely because it is rarely an issue due to the differences in mechanics between breast- and bottle feeding. (See "Neonatal oral feeding <u>difficulties due to sucking and swallowing disorders".</u>)

Infants who have problems with breastfeeding after lactation is established should be examined for ankyloglossia [6]. Consultation with a lactation consultant to exclude other causes of breastfeeding difficulty is a helpful initial step in the evaluation of an infant with ankyloglossia and difficulty breastfeeding [14]. Frenotomy may be necessary in some cases to permit effective breastfeeding [10,11,22-24]. (See "Common problems of breastfeeding and weaning" and 'Breastfeeding issues' below and 'Frenotomy' below.)

**Articulation problems** — The effect of ankyloglossia on speech is not clearly defined [2,14]. Ankyloglossia may cause articulation problems in some children, but it does not prevent vocalization or delay the onset of speech [14,25]. Frenula that extend to the tip of the tongue and prevent the tongue from reaching the upper dental alveolus are most likely to be of clinical significance. Speech sounds that may be affected include sibilants and lingual sounds (eg, "t," "d," "z," "s," "th," "n," "l"). Delayed acquisition of speech is not attributable to ankyloglossia; affected children are expected to acquire speech and language at the normal rate.

Articulation problems may not be recognized or reported by the parents [7]. Formal speech evaluation should be obtained in children who have ankyloglossia and difficulty pronouncing sibilants and lingual sounds [14]. (See "Evaluation and treatment of speech and language" disorders in children".)

**Mechanical and social problems** — Mechanical and social problems related to restricted tongue movements from ankyloglossia may include [14]:

- Difficulty with oral hygiene (ie, licking the lips or sweeping food debris from the teeth) that may result in periodontal disease (see "Gingivitis and periodontitis in children and adolescents", section on 'Gingivitis')
- Local discomfort
- Difficulty licking or kissing, which may cause social embarrassment in older children
- · Lower central incisor diastema

### **MANAGEMENT**

Consultation with a lactation specialist and/or speech pathologist may help the child to compensate for problems related to ankyloglossia, thereby avoiding surgery. Definitive treatment requires surgery.

There is a lack of consensus among clinicians regarding indications and optimal timing of surgical division for ankyloglossia. Decisions regarding surgery are usually made by the clinician(s) and the child's parents, after discussion of the potential benefits and risks of the procedure [14]. (See 'General considerations' below.)

**Breastfeeding issues** — Treatment options for infants with ankyloglossia and difficulty breastfeeding include support from a lactation consultant and/or surgical division. We suggest consultation with a lactation specialist as the first step in the management of infants with ankyloglossia and breastfeeding difficulty whose mothers wish to continue breastfeeding [14,24]. For infants who continue to have problems with breastfeeding despite lactation support, we suggest frenotomy. If frenotomy is undertaken, it should be performed by appropriately trained personnel as soon as feasible after diagnosis. [19]. (See 'Frenotomy' below.)

Frenotomy appears to have modest efficacy in improving breastfeeding outcomes in infants with ankyloglossia; however, the quality of evidence is low [26,27]. A systematic review that included five randomized clinical trials comparing frenotomy with usual care, intensive lactation consultation, or sham surgery found that frenotomy is associated with improvements in breastfeeding [26]. However, the available clinical trials were small and short term with inconsistent methodology. Four of the clinical trials reported improvements in breastfeeding using either maternally reported or observer ratings, whereas two trials found no improvement with observer ratings. Breastfeeding-associated pain scores were assessed in three trials; one reported immediate improvement in nipple pain scores among mothers of frenotomized infants, whereas the other two trials found no difference in nipple pain between the frenotomy and sham groups.

In an observational study from the province of Canterbury, New Zealand, introduction of a standardized clinical pathway for infants with ankyloglossia aimed at supporting breastfeeding and avoiding unnecessary surgery resulted in a marked decrease in the frenotomy rate (11.3 percent in 2015 to 3.5 percent in 2017), without negatively affecting the rate of breastfeeding [28].

**Articulation issues** — Formal speech evaluation should be obtained in children who have ankyloglossia and difficulty pronouncing sibilants and lingual sounds (eg, "t," "d," "z," "s," "th," "n," "l") [14]. Speech therapy may improve articulation problems in some children [14]. In the English language, even the sounds that require the most amount of tongue movement, such as "I" and "th," can be produced with minimal distortion, with the tongue tip pressed down instead of up towards the alveolar ridge ("l") or protruding out ("th"). Similarly, other sounds that require tongue elevation, such as "s" and "z," can also be produced effectively with the tongue

tip down [29]. Consequently, ankyloglossia should not have a dramatic impact on speech function in most cases [30,31]. (See "Evaluation and treatment of speech and language disorders in children", section on 'Efficacy'.)

Pediatric otolaryngologists work in concert with speech pathologists when treating children with ankyloglossia and speech problems. Often, children are referred to the otolaryngologist after a full speech assessment and a trial of therapy with a request for surgical division of the frenulum. Most otolaryngologists request input from a speech pathologist before surgery to assess the need for surgery and to document the functional nature of the procedure for thirdparty payers. Finally, speech therapy may be necessary after surgery to retrain tongue musculature and correct preoperative compensatory strategies [14].

A systematic review of studies assessing the effects of surgical treatment on articulation and speech intelligibility concluded that the evidence is insufficient to determine the benefits of frenotomy on these outcomes [31]. (See 'Surgical procedures' below.)

### Surgical procedures

**General considerations** — The goal of surgery is to increase the mobility of the tongue, not to improve its contour. Even after surgery, the tip of the tongue may retain a heart shape.

Indications for surgery may include breastfeeding difficulty, articulation problems, or mechanical/social problems [14,25]. Global speech delay is **not** an indication for surgical release of tongue-tie.

There is consensus that surgery should be performed as soon as possible after the diagnosis of ankyloglossia in an infant with breastfeeding problems not improving with conservative management [1]. The optimal timing of surgery for indications other than breastfeeding is controversial [2,3,14]. Dividing a tongue-tie in infancy has some appeal since it almost always can be accomplished without general anesthesia. Some advocate waiting until the child is at least four years old because of the chance that the frenulum may spontaneously elongate as it is used, whereas others advocate surgery before the development of feeding, speech, or other problems [14,32]. Cooperative young adults may undergo division of the frenulum under local anesthesia in the office if a tongue-tie interferes with oral hygiene or kissing.

The two most commonly performed procedures for ankyloglossia are frenotomy and frenuloplasty.

**Frenotomy** — Frenotomy (also called frenulotomy) is simple release, or "clipping," of the frenulum [14]. This procedure is often performed for infants with breastfeeding difficulty,

usually without local anesthesia [12,14,33]. (See "Prevention and treatment of neonatal pain", section on 'Local analgesia'.)

The procedure is performed under direct vision, rather than by blindly snipping the frenulum, for completeness and to avoid injury to nearby structures. The infant is restrained by swaddling or in a Papoose board, with an assistant holding the child's head. The area is illuminated with a headlight or an office microscope. The tongue is elevated with forceps, a grooved retractor, or two gloved fingers on the underside of the tongue ( picture 4). Simple division of the frenulum rarely leads to significant bleeding ( picture 5). Pressure with a gauze sponge for a few minutes controls any capillary ooze. Some surgeons prefer division of the frenulum with a carbon dioxide or diode laser, which permits bloodless incision; however, this adds additional cost to the procedure and offers no advantage in surgical results [34]. The infant may feed immediately after the procedure [14].

Potential complications of lingual frenotomy include hemorrhage, airway obstruction, injury to salivary structures, oral aversion, scarring, and recurrence. These complications are rare but do occur and should be discussed with parents during informed consent. Most studies report little discomfort and few adverse effects [10,22,35-38]. Relative contraindications to infant frenotomy include, but are not limited to, retrognathia, micrognathia, neuromuscular disorder, hypotonia, and coagulopathy [<u>1</u>].

Postoperative "tongue exercises" (eg, massaging or stretching of the tongue and floor of mouth) are used at some centers; however, the advantage of such manipulations has not been demonstrated in any clinical trial [39].

**Frenuloplasty** — Frenuloplasty is release of ankyloglossia with plastic repair. It is reserved for ankyloglossia that is not relieved by simple division of the frenulum, posterior tongue-ties, and revision cases.

Frenuloplasty requires general anesthesia, sometimes by mask but more commonly with oroor nasotracheal intubation. In this procedure, the tongue is elevated with forceps and the frenulum is divided in the sagittal plane as in frenotomy. Submucosal dissection continues into the genioglossus muscle in the midline until a full release is achieved ( <u>picture 6</u>). Bipolar electrosurgical scissors or monopolar cautery is preferred for dissection and hemostasis since the tongue musculature is vascular. Good illumination and magnification with loupes or an operating microscope permit division of the frenulum without injury to the nearby submandibular ducts. Midline dissection decreases the risks of injury to the lingual nerves.

Frenuloplasty dissection produces an elliptical tissue defect on the floor of the mouth. If left open, scarring and recurrent tethering of the tongue may result. The wound is usually closed with multiple simple absorbable sutures in the coronal plane to achieve a double V-Y advancement. Single or multiple Z-plasties have been advocated to prevent scar contracture. There are no comparative trials demonstrating an advantage to these more complicated repairs [<u>40</u>]. (See <u>"Z-plasty"</u>.)

Following surgery, a normal diet can be resumed as tolerated. In older children undergoing frenuloplasty for articulation problems, continued speech therapy and specific tongue exercises are commonly used postoperatively [41].

### SOCIETY GUIDELINE LINKS

Links to society and government-sponsored guidelines from selected countries and regions around the world are provided separately. (See "Society quideline links: Breastfeeding and infant nutrition".)

### INFORMATION FOR PATIENTS

UpToDate offers two types of patient education materials, "The Basics" and "Beyond the Basics." The Basics patient education pieces are written in plain language, at the 5<sup>th</sup> to 6<sup>th</sup> grade reading level, and they answer the four or five key questions a patient might have about a given condition. These articles are best for patients who want a general overview and who prefer short, easy-to-read materials. Beyond the Basics patient education pieces are longer, more sophisticated, and more detailed. These articles are written at the 10<sup>th</sup> to 12<sup>th</sup> grade reading level and are best for patients who want in-depth information and are comfortable with some medical jargon.

Here are the patient education articles that are relevant to this topic. We encourage you to print or email these topics to your patients. (You can also locate patient education articles on a variety of subjects by searching on "patient education" and the keyword[s] of interest.)

- Basics topic (see <u>"Patient education: Common breastfeeding problems (The Basics)"</u>)
- Beyond the Basics topic (see "Patient education: Common breastfeeding problems (Beyond the Basics)")

### SUMMARY AND RECOMMENDATIONS

- Clinical features of ankyloglossia (tongue-tie) include inability to lift the tongue to the upper <u>picture 2</u>), impaired protrusion of the tongue ( <u>picture 3</u>), impaired side-to-side movement of the tongue, and a notched or heart shape to the tongue when it is protruded ( figure 3). The natural history is unknown. (See 'Clinical features' above.)
- Potential sequelae of ankyloglossia include difficulty breastfeeding, articulation problems, and mechanical problems (eq., inability to lick the lips or sweep food debris from the teeth). Ankyloglossia does **not** cause speech or language delay. (See <u>'Potential seguelae'</u> above.)
- Indications for surgical release of ankyloglossia may include breastfeeding difficulty, articulation problems, or mechanical/social problems. The goal of surgery is to increase mobility, not to improve the contour. The optimal timing for surgery is controversial. (See 'General considerations' above.)
- For infants with ankyloglossia and breastfeeding difficulty whose mothers wish to continue breastfeeding, we suggest consultation with a lactation consultant as the first step in the management rather than surgical intervention (Grade 2C). For affected infants who continue to have problems with breastfeeding despite lactation support, we suggest frenotomy (**Grade 2C**). If frenotomy is undertaken, it should be performed by appropriately trained personnel. (See 'Breastfeeding issues' above.)
- Evaluation by a speech pathologist is an essential component of the management of children with articulation problems and concomitant ankyloglossia. (See 'Articulation issues' above.)

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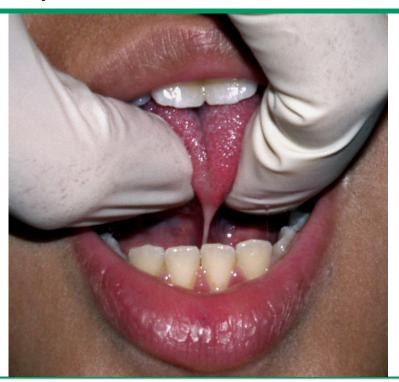
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Topic 6307 Version 24.0

### **GRAPHICS**

# Abnormally short frenulum



Abnormally short frenulum, inserting at or near the tip of the tongue.

Courtesy of Glenn C Isaacson, MD, FAAP.

Graphic 74275 Version 2.0

# Ankyloglossia in a newborn

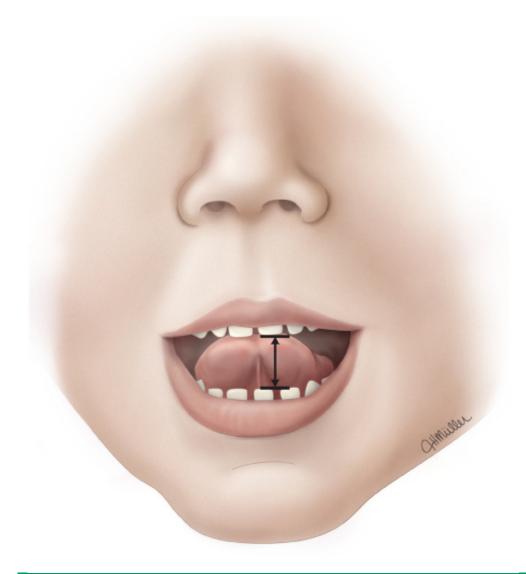


Abnormally short frenulum, inserting at the tip of the tongue in a neonate.

Courtesy of Glenn C Isaacson, MD, FAAP.

Graphic 60685 Version 2.0

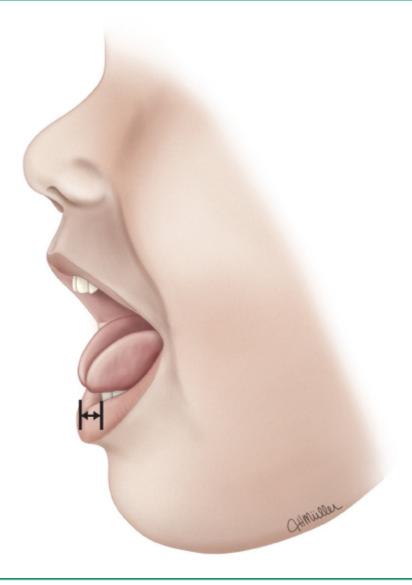
### Measurement of interincisal distance



The tongue is placed at the posterior edge of the upper teeth, and the mouth is opened as widely as possible. Measurement (in millimeters) is taken between the teeth, as illustrated above.

Graphic 72528 Version 1.0

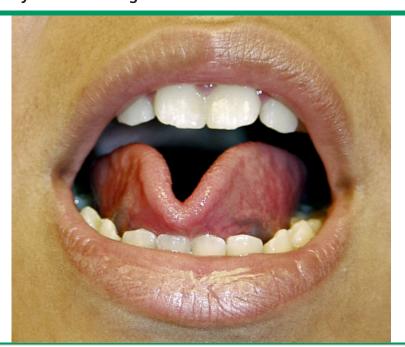
# Measurement of tongue protrusion



The tongue is protruded as far as possible. The measurement (in millimeters) is taken from the lower incisors to the tip of the tongue, as illustrated above.

Graphic 70911 Version 1.0

# Inability to lift the tongue

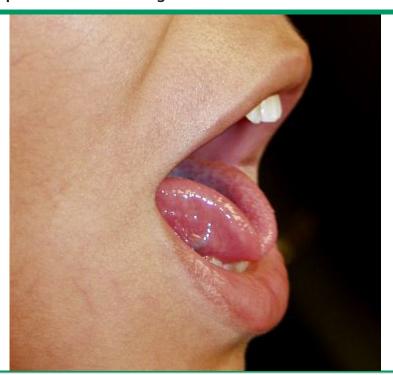


Difficulty lifting the tongue to the upper dental alveolus.

Courtesy of Glenn C Isaacson, MD, FAAP, FACS.

Graphic 50442 Version 2.0

# Short protrusion of the tongue

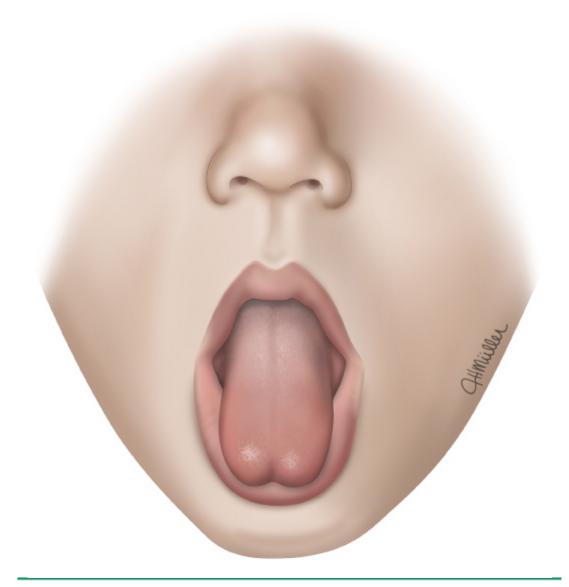


Inability to protrude the tongue more than 1 to 2 mm past the lower central incisors.

Courtesy of Glenn C Isaacson, MD, FAAP.

Graphic 61044 Version 4.0

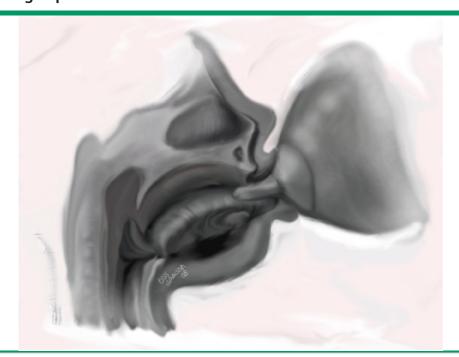
# **Notched tongue**



Notched or heart shape of the tongue when it is protruded.

Graphic 67272 Version 4.0

# **Tongue protrusion**



Protrusion of the tongue beyond the mandibular alveolus facilitates latching on during breastfeeding.

Courtesy of Tess E Isaacson.

Graphic 63201 Version 2.0

### **Frenulotomy**



The tongue is elevated with two gloved fingers to expose the lingual frenulum. The frenulum is divided close to the underside of the tongue to avoid injury to the submandibular ducts (arrows) and lingual nerves.

Courtesy of Glenn C Isaacson, MD, FAAP, FACS.

Graphic 52382 Version 1.0

# Infant status post-frenulotomy

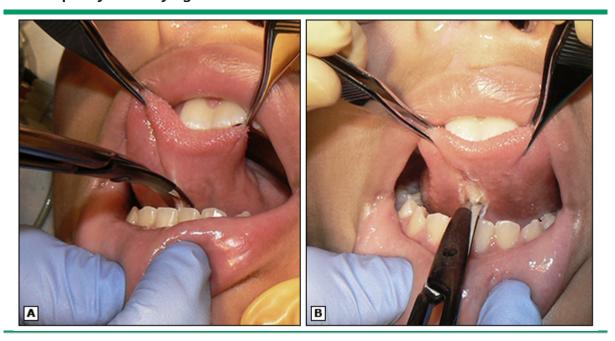


This photo shows an infant whose short frenulum has been clipped (frenulotomy).

Courtesy of Glenn C Isaacson, MD, FAAP, FACS.

Graphic 58581 Version 4.0

# Frenuloplasty for ankyloglossia



- (A) Coronal division of a tethering lingual frenulum with bipolar electrosurgical scissors.
- (B) Division of anterior fibers of the genioglossus muscle to release "posterior" ankyloglossia.

Courtesy of Glenn C Isaacson, MD, FAAP.

Graphic 53120 Version 2.0

### **Contributor Disclosures**

Glenn C Isaacson, MD, FAAP Nothing to disclose Anna H Messner, MD Nothing to disclose Carrie Armsby, MD, MPH Nothing to disclose

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