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abstract

A 6-month-old term boy was hospitalized to evaluate the cause of his failure to thrive, mandated as part of an investigation by the Department of Children and Families after an allegation of medical neglect was made. On admission the patient was below birth weight, and a medical workup for failure to thrive was pursued; however, he was noted to have severe ankyloglossia and was an exclusively breastfed infant. The only interventions during his hospitalization were frenotomy and assistance to the mother to increase her milk supply. The infant immediately experienced weight gain and has continued to show slow, but steady, weight gain as an outpatient. We illustrate here many of the controversies concerning ankyloglossia. *Pediatrics* 2010;125: e1500–e1504

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KEY WORDS

lingual frenum, failure to thrive

ABBREVIATIONS

FTT—failure to thrive

DCF—Department of Children and Families

FDRBI—frenotomy decision rule for breastfeeding infants

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FINANCIAL DISCLOSURE: The authors have indicated they have no financial relationships relevant to this article to disclose. Ankyloglossia, or tongue-tie, is a newborn condition with a reported incidence of 1% to 10%.^{1–3} Known sequelae from this condition include feeding, swallowing, and speech difficulties.^{2–5} Both the diagnosis and treatment of ankyloglossia are controversial. This case report provides an example of the consequences of untreated clinically significant ankyloglossia in an exclusively breastfed infant.

CASE REPORT

A 6-month-old white boy was admitted to the hospital for failure to thrive (FTT) after the Department of Children and Families (DCF) received an allegation of medical neglect.

This term, exclusively breastfed infant was born weighing 4.280 kg (90th percentile). His discharge weight 3 days later was 3.69 kg (14% below birth weight). While in the hospital, his mother noticed his "short tongue" and questioned providers, including a lactation consultant, about his weight loss; she was offered reassurance only. At 5 days of age, the patient's pediatrician documented a weight of 4.005 kg, and a speech-therapy evaluation for ankyloglossia was considered. His weights at 2 weeks and 2 months of age were 4.240 kg (75th percentile) and 5.510 kg (55th percentile), respectively. During the 2-week visit, complaints of maternal pain with breastfeeding and referral to oral surgery for ankyloglossia were documented. The parents reported, however, that they were told they would not find anyone to surgically intervene until at least 6 months of age and only if spoonfeeding was affected. Because of a family matter, the patient missed his 4-month well-child visit. His appointment was not rescheduled, because his parents did not think it was necessary. They expressed that they thought his growth was on track because he was "so big at birth" and he outgrew his clothes age-appropriately, unlike his premature sibling.

When the patient was 6 months old, the DCF abuse hotline received an allegation of medical neglect against his parents. The DCF instructed the parents to take the infant to their local emergency department. He was found to be hydrated and was discharged. The DCF then arranged for inpatient evaluation of FTT at the nearest teaching hospital.

On admission to our institution his weight was 4.088 kg (0.2 kg below birth weight), his length was 65 cm (10th percentile), and his head circumference was 42.5 cm (10th-25th percentile). History revealed that he breastfed exclusively for 20 minutes every 2 to 3 hours, including nighttime. His mother reported pain with breastfeeding for several weeks after birth but less discomfort at the time of admission, although he pulled back on her nipples frequently while feeding. Previous attempts at bottle-feeding were refused by the infant and, following instructions to exclusively breastfeed until 6 months of age, the parents had not yet tried supplementation with solid food. The patient's stools were of normal color and consistency and occurred every few days. Family history was significant for a 2-year-old sibling with significant growth and developmental delays secondary to premature birth at 23 weeks' gestation. Although the mother expressed milk long-term for the sibling, she never had the opportunity to breastfeed.

The patient's admission examination was significant for cachexia and severe ankyloglossia but was otherwise normal. A short, thick lingual frenulum, \sim 3 mm in height, extended the entire length of the tongue, inserting distally at the lower gingival ridge. Tongue movement was restricted to minimal bilateral elevation, with constant central depression and no extension. Aside from his severely low

weight, he had no signs of neglect. There seemed to be a strong, positive parent-infant bond. His performance on a developmental screen was closest to the level of a 5-month-old.

Initial serum laboratory work included liver-function tests, which revealed an elevated aspartate aminotransferase (AST) level of 123 U/L (normal: 15-60 U/L) and an alanine aminotransferase (ALT) level of 83 U/L (normal: 13-45 U/L) but were otherwise normal. His prealbumin level was <6 mg/dL (normal: 20-40 mg/dL). His complete blood count results were as follows: white blood cell count, $5700/\mu$ L (normal: 6000-17 500/µL), with 19% neutrophils (normal: 35%-85%), 74% lymphocytes (normal: 25%-65%), 4% monocytes (normal: 2%-10%), and 3% basophils (normal: 0%-2%); hematocrit, 27.7% (normal: 29%-41%); and platelet count, $321\,000/\mu$ L (normal: $150\,000-450\,000/\mu$ L). His thyroidfunction test results were slightly abnormal, with a free thyroxine level of 0.81 ng/dL (normal: 0.93-1.7 ng/dL), triiodothyronine level of 75 ng/dL (80-200 ng/dL), and thyrotropin level of 4.36 mIU/L (normal: 0.27-4.2 mIU/L). Results of a urinalysis and basic metabolic panel were unremarkable.

The initial plan included interruption of breastfeeding, bottle-feeding only with expressed breast milk or formula, strict intake documentation, nutrition consultation, and possible further metabolic and gastrointestinal workup if indicated. The attending physician modified this plan by having the mother resume breastfeeding and requesting a breastfeeding medicine consult. The breastfeeding medicine team, which consisted of a neonatologist with international board-certified lactation-consultant status and an occupational therapist, recommended frenotomy, prebreastfeeding and postbreastfeeding weight measurements, attempting supplementation by using

a supplemental nurser system, and increasing maternal milk supply (express milk after each feed, continue fenugreek). The patient refused intake via bottle, supplemental nurser system, and syringe feeding. Frenotomy was performed on hospital day 1. The next day he experienced no weight change, and his physical examination still showed restricted tongue movement, so the frenotomy incision was extended. His mother noted marked improvement in pain and stated that "it feels like there's a pillow under there." Milk transfer, according to prefeeding and postfeeding weights, increased from 5 mL before frenotomy to 56 mL immediately afterward and to a maximum of 190 mL after extension of the frenotomy. His weight on hospital day 3 was 4.760 kg. On hospital day 4 his weight was 5.040 kg (up 0.952 kg from admission), at which time he was discharged from the hospital. FTT was attributed to feeding problems and inadequate intake as a result of ankyloglossia. Despite unequivocal statements in the hospital records showing a purely anatomic cause for the patient's FTT, the investigation by the DCF remained open.

DISCUSSION

Ankyloglossia is a relatively common newborn condition for which the diagnosis and treatment are controversial. The Academy of Breastfeeding Medicine defines ankyloglossia as "the presence of a sublingual frenulum which changes the appearance and/or function of the tongue because of decreased length, lack of elasticity, or attachment too distal beneath the tongue or too close to the gingival ridge."⁶ Ankyloglossia has been associated with feeding, swallowing, and speech difficulties²⁻⁵ and is particularly important in breastfeeding infants, in whom it has been associated with poor latch, maternal nipple pain, poor milk transfer, and FTT.⁷ Objective

Mothers name: Assessment Tool for Lingual Frenulum Baby's name: Function (ATLFF) Baby's age: © Alison K. Hazelbaker, PhD, IBCLC March 1, 2009 Date of assessment: FUNCTION ITEMS Lateralization Cupping of tongue 2 Complete 2 Entire edge, firm cup 1 Body of tongue but not tongue tip 1 Side edges only, moderate cup 0 None 0 Poor OR no cup Lift of tongue Peristalsis 2 Tip to mid-mouth 2 Complete anterior to posterior (originates at tip) 1 Only edges to mid mouth 1 Partial: originating posterior to tip 0 Tip stays at alveolar ridge OR tip rises only 0 None OR Reverse peristalsis to mid-mouth with jaw closure AND/OR midtongue dimples Extension of tongue Snap back 2 Tip over lower lip 2 None 1 Tip over lower gum only 1 Periodic 0 Neither of the above OR anterior or mid-0 Frequent OR with each suck tongue humps and/or dimples Spread of anterior tongue 2 Complete 1 Moderate OR partial 0 Little OR none **APPEARANCE ITEMS** Elasticity of lingual frenulum Appearance of tongue when lifted 2 Round **OR** square 2 Very elastic (excellent) 1 Slight cleft in tip apparent 1 Moderately elastic 0 Heart shaped 0 Little OR no elasticity Length of lingual frenulum when tongue Attachment of lingual frenulum to tongue lifted 2 Posterior to tip 2 More than 1 cm OR absent frenulum 1 1 cm 1 At tip 0 Notched **OR** under the mucosa at the tongue 0 Less than 1 cm base Attachment of lingual frenulum to inferior alveolar ridge Attached to floor of mouth OR well below ridge 1 Attached just below ridge 0 Attached to ridge SCORING Function Item score: Combined Score: Appearance Item score: **Treatment Recommendations Based on** Scoring 14 = Perfect Function score regardless of Appearance Item score. Surgical treatment not recommended. 11 = Acceptable Function score only if Appearance Item score is 10. <11 = Function Score indicates function impaired. Frenotomy should be considered if management fails. Frenotomy necessary if Appearance Item score is < 8.

 TABLE 1
 Hazelbaker Assessment Tool for Lingual Frenulum Function

(Reproduced with permission from Hazelbaker AK. *The Assessment Tool for Lingual Frenulum Function (ATLFF): Use in a Lactation Consultant Private Practice* [master's thesis]. Pasadena, CA: Pacific Oaks College; 1993:64–65.)

assessment of ankyloglossia may use the Hazelbaker scale (Table 1), which quantifies aspects of lingual appearance and function,⁸ or the frenotomy decision rule for breastfeeding infants (FDRBI) (Table 2), which uses signs and symptoms.⁴

The preferred treatment for clinically significant ankyloglossia is frenotomy, which involves simple incision of the lingual frenulum with surgical scissors.^{2,6} Although the therapeutic utility of frenotomy has been debated, results of multiple studies have shown clear evidence that frenotomy improves breastfeeding in mother-infant dyads who are experiencing breastfeeding difficulties.^{2,4,5,7} Geddes et al² showed that for breastfeeding infants who were experiencing feeding problems, frenotomy improved tongue movement and nipple distortion, as imaged by ultrasound, and improved milk intake, milk transfer, attachment, and maternal nipple pain. Results of 2 prospective randomized, controlled trials in infants with breastfeeding problems showed frenotomy to be a safe and effective therapy,^{5,9} and authors of another study reported improved latch scores and decreased maternal pain after frenotomy.⁴

A survey study evaluated the opinions and practice regarding ankyloglossia among otolaryngologists, pediatricians, lactation consultants, and speech pathologists and revealed no consensus regarding the significance of ankyloglossia or indications for treatment,¹ probably related to the lack of an accepted standard for diagnosis.^{1,2,4,6,7,10} Although both the Hazelbaker assessment and the FDRBI have been proposed as standardized tools, neither has gained widespread acceptance.^{4,8} One advantage of the FDRBI is that it incorporates maternal signs and symptoms, thereby adding functional assessment to the anatomic one. How mother and infant fit and perform together is the critical factor, not either one in isolation. The authors asserted, and FDRBI supports, that if ankyloglossia and breastfeeding problems coexist and cannot be easily remedied by correction of latch or

TABLE 2	Frenotomy [Decision	Rule for	Breastfeeding Infants	
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a visible

Mother with nipple pain or trauma while breastfeeding				
AND/OR				
inability to maintain latch				
AND/OR				
poor weight gain in the infant ($\leq 15 \text{ g/d}$)				
AND				
e membrane anterior to the base of the tongue, which restricts tongue movement, leading to:				
inability to touch the roof of the mouth				
OR				
inability to cup an examining finger				
OR				

inability to protrude the tongue past the gum line

(Reproduced with permission from Srinivasan A, Dobrich C, Mitnick H, Feldman P. *Breastfeed Med.* 2006;1[4]:218.)

other typical lactation interventions, then frenotomy is indicated.⁴

Feeding concerns, including breast pain, poor latch, and incomplete milk transfer, were raised by our patient's mother during his newborn hospital stay and at his first 2 well-child visits. Despite these concerns, reassurance was the only "intervention" provided by multiple health care providers. The option of referral to an oral surgeon was offered but discouraged. Yet, over the first 2 months of his life the patient crossed 3 major percentile markers on his weight growth curve. Admittedly, his parents had a distorted perception of normal growth and development that was based on their experience with the patient's extremely premature sister. However, he was growing into larger-sized clothing and his development appeared acceptable, so his malnutrition was not readily evident. On admission, the patient was below birth weight at 6 months of age and had an undetectable prealbumin level. After frenotomy and increasing maternal milk supply, he immediately experienced weight gain, and his mother reported improved latch and resolution of breast pain. Since discharge the patient has continued to show slow, steady weight gain and maintain a curve below the 3rd percentile. Despite multiple occasions on which the mother expressed concerns about breastfeeding and ankyloglossia, no interventions occurred until he reached a state of severe malnutrition. By continuing to exclusively breastfeed, as recommended by her pediatrician and the American Academy of Pediatrics,¹¹ this infant starved and his mother was accused of neglect. Although the DCF closed its investigation after 6 months, this family now has a record with the DCF and must endure the associated stigma and connotations.

CONCLUSIONS

This experience illustrates how crucial tongue mobility is for successful breastfeeding and how important it is to take seriously maternal concerns regarding breastfeeding pain and difficulty. Our mandate as pediatricians is to maximize breastfeed-

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ing; therefore, we are obliged to identify and rectify risk factors for breastfeeding failure and to be aware of the consequences of failing to do so. By promoting exclusive breastfeeding to those who are at risk for failure and then ignoring risk factors, are we not the negligent

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parties? The ramifications of treatment decisions associated with ankyloglossia are complex and may seriously affect physical, mental, and social health. This example of a breakdown in our medical system resulted in infant morbidity and a false accusation of child abuse.

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