### **ARTICLE IN PRESS**

International Journal of Pediatric Otorhinolaryngology xxx (2010) xxx-xxx



Contents lists available at ScienceDirect

### International Journal of Pediatric Otorhinolaryngology

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



### Defining ankyloglossia: A case series of anterior and posterior tongue ties

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#### ARTICLE INFO

Article history: Received 18 March 2010 Received in revised form 18 May 2010 Accepted 23 May 2010 Available online xxx

Keywords: Ankyloglossia Posterior Ankyloglossia Frenotomy Tongue tie release Breastfeeding difficulties

#### ABSTRACT

*Introduction:* Ankyloglossia is a congenital condition in which tongue mobility is limited due to an abnormality of the lingual frenulum. The impact of ankyloglossia on breastfeeding is poorly understood but there is a recent trend toward more recognition of this condition and early intervention when needed. Currently, there lacks clear definition of ankyloglossia and different subtypes have been proposed with no clinical correlation.

*Objective:* To determine the prevalence of anterior versus posterior ankyloglossia in a large series of consecutive patients and to assess clinical outcomes after frenotomy.

*Methods:* Retrospective chart review of patients from July 2007 to July 2009 who were diagnosed with ankyloglossia and underwent office frenotomy. Baseline characteristics, specific feeding issues, type of ankyloglossia, and clinical outcomes after frenotomy were reviewed.

*Results:* Of the 341 total patients, 322 (94%) had anterior ankyloglossia and 19 (6%) had posterior ankyloglossia. Median age at presentation was 2.7 weeks (range 1 day of life to 24 weeks); 227 were males and 114 were females. Revision frenotomy rates were significantly higher for the posterior ankyloglossia group (3.7% anterior and 21.1% posterior, p = 0.008).

*Conclusion:* Anterior ankyloglossia is much more common and readily managed when compared to posterior ankyloglossia. Posterior ankyloglossia is a poorly recognized condition that may contribute to breastfeeding difficulties. The diagnosis is difficult due to the subtle clinical findings but relevant health care providers should be aware of this condition. Frenotomy is a simple, safe, and effective intervention for ankyloglossia which improves breastfeeding.

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#### 1. Introduction

Ankyloglossia or tongue tie refers to a congenital problem characterized by an abnormal lingual frenulum which can limit tongue mobility. In the past, ankyloglossia was thought to significantly impact breastfeeding and thus frenotomy (tongue tie release) procedures were commonly performed in infants [1]. The increased availability and popularity of formula and bottle feeding reduced the focus on breastfeeding and thus the significance of ankyloglossia. However, with the recognition that breastfeeding confers many advantages to both infants and mothers, there is an increased expectation that mothers will breastfeed their newborn infants [2].

Traditionally, the impact of ankyloglossia on breastfeeding was unclear. Couple of articles have stated that an infantile ankyloglossia does not contribute to breastfeeding problems [1,3]. This is in keeping with the fact that many infants with ankyloglossia can breastfeed satisfactorily but the assertion that all, or most,

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newborns are unaffected is not evidence based. There are recent studies suggesting an association between ankyloglossia and breastfeeding difficulties [4–7] and resolution of breastfeeding issues with frenotomy procedures [8–11]. Some physicians and many lactation consultants now believe that ankyloglossia can make breastfeeding difficult resulting in sore nipples and in some instances mastitis or other painful conditions for the mother, as well as being associated with poor weight gain and early weaning in some infants [8,12,13].

Currently, there is no consensus regarding the precise definition of ankyloglossia and while several classification systems have been proposed to grade the degree of ankyloglossia [5,13–15], none of the systems have been correlated to symptomatic severity. Some clinicians have defined ankyloglossia according to the anterior position of the lingual frenulum and the associated tongue tip movement restriction [10,13,16]. However, the more subtle posterior ankyloglossia is rarely reported to cause any difficulties with breastfeeding. There is only one case report describing posterior ankyloglossia and feeding problems known to these authors [17].

We present a case series of infants who underwent outpatient frenotomy for breastfeeding difficulties associated with anterior

Please cite this article in press as: P. Hong, et al., Defining ankyloglossia: A case series of anterior and posterior tongue ties, Int. J. Pediatr. Otorhinolaryngol. (2010), doi:10.1016/j.ijporl.2010.05.025

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and posterior ankyloglossia. Clinical presentations, physical findings, specific indications and outcomes of frenotomy are described.

### 2. Patients and methods

Retrospective chart review of patients seen in an outpatient pediatric otolaryngology clinic for ankyloglossia was carried out. Subjects were identified using the ICD.9 code for ankyloglossia and the CPT code for outpatient frenotomy from July 2007 to July 2009. Patient demographics, presenting complaints, and clinical outcomes were recorded. The study population composed of healthy infants with no other significant medical issues.

All patients underwent full head and neck examination by the attending clinicians which included palpation of the floor of mouth and lingual frenulum. Ankyloglossia, if present, was then classified as either anterior or posterior. The grading was subjectively determined by the examiner based on the physical prominence, tightness, and location of the lingual frenulum on inspection and palpation, as well as the apparent limitation of tongue protrusion and notching of the tongue tip.

Office frenotomy was performed in infants younger than 24 weeks of age. All patients had difficulties with breastfeeding and all mothers still desired to breastfeed. Some presented with persistent feeding issues despite professional help from lactation consultants. More specifically, these problems included difficulties latching onto the breast by the infant, sore nipples experienced by the mother, prolonged feeding times, and poor weight gain.

All frenotomy procedures were performed after obtaining informed consent in an outpatient clinic using a standard technique which included the application of topical anesthetic agent (20% Benzocaine). First, the grooved director was used to retract the ventral tongue to fully expose the lingual frenulum. A straight hemostat was then clamped parallel to the tongue at the ventral attachment of the frenulum; after waiting few seconds, the clamp was released and sterile iris scissors was used to release the lingual frenulum. Care was taken not to injure the submandibular ducts. After the division, the floor of mouth was compressed with gauze to provide hemostasis. The infant was immediately returned to the mother to be nursed. All patients were asked to contact the clinic if there were any persistent issues with breastfeeding.

All patients tolerated the procedure well and there were no reported complications.

#### 3. Results

A total of 341 patients were identified as having the diagnosis of ankyloglossia and underwent outpatient frenotomy during the study period. Two-hundred and twenty seven subjects were males and 114 were females. Median age at presentation was 2.7 weeks (range 1 day of life to 24 weeks).

Of the 341 patients, 322 (94%) had anterior ankyloglossia and 19 (6%) were classified as having posterior ankyloglossia. Anterior ankyloglossia was defined as tongue ties with a prominent lingual frenulum and/or restricted tongue protrusion with tongue tip tethering. The diagnosis of posterior ankyloglossia was considered when the lingual frenulum was not very prominent on inspection but was thought to be tight on manual palpation or was found to be abnormally prominent, short, thick or fibrous cord-like with the use of the grooved director (Figs. 1 and 2). All patients with both anterior and posterior ankyloglossia had significant breastfeeding problems on history.

There was a male predominance in patients with anterior tongue ties while the converse was observed for posterior ankyloglossia, which showed a larger female predominance (Table 1). Age at presentation was similar for both groups while



Fig. 1. Posterior ankyloglossia on inspection (please note that this photo was taken in the operating room on a non-study older patient).



Fig. 2. Posterior ankyloglossia with retraction of ventral tongue with a grooved director (same patient as in Fig. 1). Lingual frenulum is much more prominent with retraction

the rate of revision frenotomy required was significantly different (3.7% versus 21.1%, p = 0.008). Patients with posterior ankyloglossia were much more likely to need revision frenotomy when compared to patients with anterior ankyloglossia. All patients in both groups did well after the revision procedures with improved breastfeeding.

Table 2 shows the specific breastfeeding issues for the group of infants with posterior ankyloglossia. Most common presenting complaint was maternal nipple pain (89%). Maternal nipple pain was described by the mothers as sore nipples during and after breastfeeding. All mothers with nipple pain also complained of bloody nipple discharge, presumably from ankyloglossia associated nipple trauma. The complaint of maternal nipple pain was followed closely by latching-on difficulties (84%) and prolonged feeds (80%). The specific definition of prolonged feeds (length of time per feed or amount of breastmilk in certain amount of time)

#### Table 1

Patient baseline characteristics, types of ankyloglossia, and revision rates of tongue tie release.

	Anterior (N=322)	Posterior (N=19)	p Value
Gender			
Female	101 (31.4%)	12 (63.2%)	
Male	221 (68.6%)	7 (36.8%)	
Age at presentation (weeks)	2.4	2.9	0.5
Revision procedures	12 (3.7%)	4 (21.1%)	0.008

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#### Table 2

Characteristics of patients with posterior ankyloglossia.

	Latching issues <sup>a</sup>	Poor weight gain <sup>a</sup>	Maternal nipple pain <sup>a</sup>	Prolonged feeds <sup>a</sup>	Lactation consultation <sup>a</sup>	Provider <sup>b</sup>	Revision <sup>a</sup>
1	Y	Ν	Y	Ν	Y	DL	
2	Y	Ν	Y	Y	Y	AM	
3	Y	Ν	Y	Y	Y	SMP	
4	Y	Ν	Ν	Ν	Ν	SMP	
5	Y	Ν	Y	Y	Y	SMP	
6	Y	Ν	Y	Ν	Y	AM	
7	Y	Ν	Y	Ν	Y	AM	Y
8	Y	Ν	Y	Y	Y	SMP	
9	Y	Ν	Ν	Y	Ν	AM	
10	Y	Ν	Y	Y	Ν	AM	
11	N	Ν	Y	Y	Ν	AM	Y
12	Y	Ν	Y	Y	Y	DL	Y
13	Ν	Ν	Y	Y	Y	DL	
14	Y	Y	Y	Y	Y	DL	
15	Y	Ν	Y	Y	Y	SMP	
16	Y	Y	Y	Y	Y	SMP	
17	Y	Y	Y	Y	Y	SMP	
18	Ν	Ν	Y	Y	Y	DL	Y
19	Y	Ν	Y	Y	Υ	SMP	

<sup>a</sup> Y=yes; N=no.

<sup>b</sup> Author/provider initials.

was not consistently documented in the patient charts and thus we could not further detail this presenting complaint. Poor weight gain was seen in 3 patients (16%). Lactation consultants were involved in majority of the cases (80%) and all of these patients showed no significant improvements with conservative intervention alone. Conservative intervention included professional assistance from lactation consultants, which mainly involved trials with different positioning techniques for up to 1 week.

#### 4. Discussion

Recent studies suggest that there is an association between breastfeeding difficulties and ankyloglossia [4–7] and some articles have suggested favorable outcomes after frenotomy in symptomatic infants with ankyloglossia [8–11]. These findings, along with the dramatic increase in lactation consultancy have significantly increased the awareness of this treatable cause of failure to breastfeed.

The incidence of tongue tie is estimated to range from 0.02% to 4.8% [4,18,19] with a male predominance (male to female ratio of 2.6:1.0) [20]. The wide variation of the incidence may stem from a lack of uniform definition and objective classification system.

Several studies have tried to establish a criteria for diagnosing ankyloglossia: the diagnosis has been based on the length of the lingual frenulum [4,13,14,18], the amount of tongue movement [12,16,21,22], the appearance of the tongue tip being 'heartshaped' [10], and on thick fibrous cord being palpated on physical examination [17,18]. In the lactation literature ankyloglossia is defined on a more general and subjective basis, mainly considering the symptomatic complaints of the mother [8]. Our experience suggests that the classic definition of ankyloglossia which includes the lingual frenulum being prominent or short is not always accurate as some infants with subtle posterior ankyloglossia can have breastfeeding problems. Clearly, there needs to be a uniform definition which combines both the physical examination findings and the historical aspects of breastfeeding problems that can be easily adopted by all involved health care professionals.

Currently, there is no commonly applied grading system for ankyloglossia in the clinical setting. Many parents will describe the anterior ankyloglossia with tongue tip tethering as 'heart-shaped' but other less prominent types are not described with any consistency. Some have proposed a classification system based on the amount of tongue protrusion, ranging from 0% to 100% [16], while others also include the thickness of the lingual frenulum [15]. As demonstrated by our case series of patients with posterior ankyloglossia and another large study [4], there does not seem to be a clear correlation between the grade and severity of ankyloglossia and breastfeeding difficulties. These findings suggest that the degree of clinical manifestation is not clearly associated with the physical appearance of the lingual frenulum and breastfeeding problems can arise in anterior or posterior ankyloglossia.

When indicated, tongue tie release or frenotomy is a quick and simple procedure that can be accomplished in an office setting at the time of the initial consultation. Typically, the discomfort for the infants is minimal and they are allowed to breastfeed immediately after the procedure. The infantile lingual frenulum tends to be thin and relatively avascular, resulting in minimal bleeding after the division. Potential complications include damage to the tongue and submandibular duct and orifice but they are extremely rare [10].

There are several studies that have assessed the issue of whether failure to breastfeed in an infant with ankyloglossia can be helped by simple division of the lingual frenulum and several studies advocate frenotomy in symptomatic infants [8–11]. This is also evident in our experience and mothers frequently report an immediate subjective improvement in breastfeeding mechanics. Moreover, lactation consultants routinely report dramatic improvements in nursing after frenotomy.

Infants with posterior ankyloglossia and breastfeeding problems underwent office frenotomy without any significant complications in our series. When compared to anterior subtypes, there were significantly higher rates of revision procedures (3.7% anterior versus 21.1% posterior). This may be explained by the subtle and extreme posterior nature of the lingual frenulum. Revision cases were carried out in the standard fashion, ensuring that the posterior most frenulum was divided. Adequate frenotomy was verified by observing the diamond-shaped cut mucosa with visualization of the underlying musculature. With this technique, all revision cases reported improved breastfeeding.

This study is far from conclusive as there were only 19 patients in the posterior tongue tie group. Other potential shortcomings include the retrospective nature of the study, absence of specific definitions of presenting complaints (length of 'prolonged feeds') and the lack of long term follow-up. As well, the post-procedure outcome assessment was not specific and we did not evaluate the exact length or quality of the nursing experiences. The determina-

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tion of anterior versus posterior ankyloglossia was subjective and may have resulted in some infants being misclassified. Finally, the present study addresses the need for intervention as it relates only to breastfeeding issues. We do not attempt to study the other ankyloglossia associated morbidity including anticipated speech and social issues.

#### 5. Conclusion

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Anterior ankyloglossia is much more common and readily managed when compared to posterior ankyloglossia. The anterior subtype is more promptly recognized by health care professionals while posterior ankyloglossia is a relatively unknown and rarely recognized condition that may be associated with breastfeeding difficulties. Posterior ankyloglossia may often be missed due to its subtle nature and it may require palpation or exposure with a grooved director for identification. Office frenotomy is a safe and effective method that can provide immediate symptom relief and promote breastfeeding. The need for revision frenotomy is higher for posterior ankyloglossia when compared to anterior ankyloglossia, however, with diligent technique mothers can breastfeed successfully.

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