



Parental perception of speech and tongue mobility in three-year olds after neonatal frenotomy



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ABSTRACT

Objectives: The goal of this study was to evaluate parental speech outcomes and tongue mobility in children with ankyloglossia who underwent frenotomy by an otolaryngologist during the neonatal period.

Study design: Cohort study and retrospective telephone survey.

Study setting: University Hospital.

Subjects and methods: Neonates previously diagnosed with congenital ankyloglossia were separated into Surgical Intervention ($N = 71$) and No Surgical Intervention ($N = 15$) Groups. A Control Group ($N = 18$) of patients was identified from the hospital medical record database, which were not diagnosed with congenital ankyloglossia. A survey provided by a certified speech pathologist utilized a Likert scale to assess speech perception and tongue mobility by parental listeners. The questionnaire also analyzed oral motor activities and the medical professionals that identified the ankyloglossia shortly after birth. Statistical analyses were performed with the Wilcoxon Rank Sum Test and Fischer's Exact Test in order to determine an effect size = 1.

Results: There was significantly improved speech outcomes designated by parents in the Surgical Intervention Group when compared to the No Surgical Intervention Group [$p < 0.0001$, $p < 0.0001$], respectively. Furthermore, parents designated no difference in speech outcomes between the Surgical Intervention Group when analyzed against the Control Group [$p = 0.3781$, $p < 0.2499$], respectively.

Conclusions: There was a statistically significant improvement in speech outcomes and tongue mobility in children who underwent frenotomy compared to individuals who declined the operation. As a result of the data presented within this study, there appears to be a long-term benefit beyond feeding when frenotomy is performed in newborns with ankyloglossia.

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1. Introduction

Ankyloglossia, commonly known as tongue-tie, is a congenital anomaly characterized by a short or tight lingual frenulum, which results in limited tongue movement [1]. Incidence rates vary from 2 to 10% with a male predominance of about 3:1 and several classification systems have been proposed but none have gained universal acceptance [2,3]. Currently there is controversy regarding the significance, diagnosis, natural history and appropriate

management of ankyloglossia amongst pediatricians and specialists [4].

Tongue-tie division (frenotomy/frenulectomy or frenuloplasty) has been performed since at least the 18th century and perhaps longer [1]. Recently, several publications have elucidated the benefits of frenotomy with breastfeeding when performed on newborns including infant weight gain, reduced maternal pain during nursing, improved latch and reduced compression of the nipple [5–8]. Several subjective indicators regarding increased maternal satisfaction and diminished anxiety were described as well [3,9].

While the advantages of this procedure are well documented with regard to breastfeeding, currently, the relationship between frenotomy and speech outcomes is a lightly investigated topic. To the best of our knowledge there have only been four original

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studies evaluating speech after lingual frenulum division which are limited by either small population size, lack of control subjects or both [10–13]. Messner and Lalakea demonstrated improved speech articulation in both children and adults in two separate studies while Dollberg et al. were not able to definitively conclude a benefit [10–12]. Heller et al. showed a speech benefit from a Z-frenuloplasty but not from a traditional frenotomy [13]. Statistical significance was limited in all of these studies due to the small size and lack of control subjects, which underscores the current controversy and dearth of information on the effect of frenotomy on speech. In this study, we retrospectively compared a large cohort of three-year old patients who underwent frenotomy for ankyloglossia as neonates with their untreated counterparts and a healthy group without ankyloglossia as a control. To date this is the largest evaluation of speech outcomes in patients that received frenotomy shortly after birth.

2. Methods

2.1. Patient selection

The study was approved through the Georgetown University Institutional Review Board and informed consent was obtained from family members before the initiation of the telephone survey. The Surgical Intervention Group was assembled from the medical records of 102 three-year old patients with a past history of ankyloglossia who had received frenotomy within the first month of life between January 2010 and December 2010. The No Surgical Intervention Group was obtained from the medical records of 21 three-year old patients with a past history of ankyloglossia but declined the frenotomy procedure during the same time period. The Control Group was compiled from the medical records of randomly assigned three-year old patients with no past history of ankyloglossia during the same time period. The degree of ankyloglossia was determined by a pediatric otolaryngologist in the post-partum ward or during outpatient clinical examination.

Patients were then cross-referenced to blind the interviewer and the statistician. The study age was chosen after consultation with a certified speech pathologist and review of the literature demonstrated that three years of age was when the majority of children begin to develop speech and articulation abnormalities if present [14,15].

Within these cohorts, patient information was compiled regarding: age, gender, degree of ankyloglossia, family history of ankyloglossia and if the frenotomy was performed in the otolaryngology clinic or postpartum ward. A grading scale ranked the tongue-tie from types 1 to 4 based on the position of the frenulum by using the Coryllos criteria [16].

2.2. Telephone survey

The telephone survey was prepared by a certified speech pathologist and consisted of nine questions regarding the healthcare provider who identified the restricted lingual frenulum, the healthcare provider who recommended the surgery, intelligibility of speech to a mother or father, impaired speech sounds, deficiencies in oral motor activities, and the perceived need for speech therapy. Quantitative data regarding speech intelligibility was compiled by utilizing the Likert Scale 1–5 (1 – Poor outcome, 3 – Intelligible, 5 – Well-developed). Parents were asked to consider vocabulary development, articulation and impaired sounds when making their judgments regarding speech outcomes (Fig. 1).

2.3. Statistical analysis

The research question of interest is to compare the speech outcomes in children with tongue-tie/ankyloglossia who underwent

Table 1
Study group and ankyloglossia type distribution.

Study group and ankyloglossia type		(%)
Total number of patients	104	(100)
Surgical Intervention Group	71	(69)
No Surgical Intervention Group	15	(14)
Control Group	18	(17)
Gender		
Male	62	(59)
Female	42	(41)
Mean age of patients (days)	9	
Family history of patients with ankyloglossia		
Yes	27	(32)
No	59	(68)
Type of Ankyloglossia		
1	20	(23)
2	44	(51)
3	22	(26)
4	0	(0)

frenotomy to those who did not during the neonatal period, using the survey questions that employed a Likert scale to assess speech perception by parents. Since there is no available published study of such kind, we planned to have 60 children in the frenotomy group and 15 children in the non-frenotomy group, which would provide at least 90% power to detect an effect size = 1, based on a two-sided test with a significant level of 0.05.

Descriptive statistics (e.g., mean, median, proportion) were used for patients' demographic information. Fisher's Exact test was used to compare the categorical variables. Since the speech perception is based on a 1–5 scale, the distribution of the score is not normally distributed. The Wilcoxon Rank sum test was utilized to compare children's speech outcomes as described by parents.

3. Results

A total of 104 children were included within this study and the mean age was nine days (Table 1). Hospital lactation consultants most frequently identified the patient's ankyloglossia and more frequently recommended the frenotomy procedure when compared to pediatricians, parents and other healthcare providers (Table 2).

Of the 86 patients that were included in the Surgical Intervention and No Surgical Intervention group, parents reported 36 to have some form of difficulty with speech. Mean speech outcomes of the Surgical Intervention, No Surgical Intervention and Control Groups were evaluated by parental listeners via telephone survey. Parents designated significantly improved speech outcomes when Surgical Intervention Group patients were compared to No Surgical Intervention Group patients, respectively. Furthermore, listeners reported no differences in speech outcomes between the Surgical Intervention and the Control Groups, respectively (Tables 3 and 4). In addition to the improved speech outcomes, parents reported significant improvements in their child's ability to clean the vestibule of the mouth, clean the outside of their mouth with their tongue and consume ice cream (Table 5).

Table 2
Percentage of healthcare providers identifying tongue-tie and recommending frenotomy.

Healthcare provider	Percent of tongue-ties identified (%)	Percent of frenotomies recommended (%)
Lactation consultant	64	55
Pediatrician	22	33
Other (NP, OB)	16	12

1. Who identified the tongue-tie and what was the reasoning for evaluation (e.g., parent/doctor; child having difficulty or noted on routine exam)?
2. Was it recommended to you to have your child’s tongue clipped? By whom?
3. Did you have your child’s tongue-tie clipped (If asked of a control, and responds yes, then patient can no longer be used as a control)?
4. On a scale of 1-5 how would you rate your child’s speech outcome now? Please consider vocabulary, articulation and sentence structure when deciding.

1	2	3	4	5
Poor		Intelligible		Well - Developed
5. What, if any, speech sounds would you report are impaired?
6. Does your child have difficulty with any of the following oral-motor activities?
 - a. Cleaning the inside of their mouth?
 - b. Licking outside of lips?
 - c. Eating ice cream?
 - d. Touching tongue to upper lip?
7. Were you recommended to or did you seek out speech therapy services? If so, how was your child’s speech to a familiar listener prior to intervention by speech therapy?

1	2	3	4	5
Poor		Intelligible		Well - Developed
8. Were you recommended to or did you seek out speech therapy services? If so, how was your child’s speech to a familiar listener after speech therapy?

1	2	3	4	5
Poor		Intelligible		Well - Developed
9. If you did not receive a frenulectomy, do you feel that your child needs speech therapy?

Fig. 1. Administered telephone speech survey.

Table 3
Likert scale speech outcomes survey results (1 – Poor outcome, 3 – Intelligible, 5 – Well-developed).

	Surgical Intervention	No Surgical Intervention	Control
Mean (SD) speech outcome	4.52 (0.61)	3.60 (0.63)	4.33 (0.77)

Table 4
Wilcoxon rank sum test results of speech outcomes.

Parental speech evaluation	Speech outcome comparison	p-Value
	Surgical Intervention vs. No Surgical Intervention	<0.0001
	Surgical Intervention vs. Control	0.3781
	No Surgical Intervention vs. Control	0.01

4. Discussion

Ankyloglossia affects approximately 2–10% of newborns and is routinely treated in outpatient centers when mothers report breastfeeding difficulties [2,3,17]. Overall, frenotomy seems to be an effective and quick method to alleviate breastfeeding difficulties in patients with ankyloglossia, but little information is present in the medical literature regarding its long-term effects on speech

outcomes. The current study provides the first large-scale analysis of the long-term effects of frenotomy on speech outcomes.

Currently, the clinical benefits of frenotomy remain a hotly debated topic between pediatricians, otolaryngologists and lactation consultants. In many cases, pediatricians and a small subgroup of otolaryngologists question the potential benefit of frenotomy and this is elucidated in an article published by Ballard et al. A survey of 831 healthcare providers reported that pediatricians were most likely to recommend against having frenotomy performed in the newborn stage while lactation consultants were the primary impetus for proposing Surgical Intervention [17]. This finding parallels the results obtained in our study, as lactation

Table 5
Fischer’s exact test p-values regarding improved oral motor activities after frenotomy.

	Surgical Intervention, vs. No Surgical Intervention	Surgical Intervention vs. Control	No Surgical Intervention vs. Control
Difficulty cleaning teeth with tongue	0.0006	1.0000	0.0120
Difficulty licking outside of lips	<0.0001	0.1120	0.0053
Difficulty eating ice cream	0.0003	0.5786	0.0015

consultants identified and recommended more than half of our parents to an otolaryngologist for frenotomy (Table 2). Interestingly, survey responses indicated that the lactation consultant recommended the operation in order to alleviate difficulties with breastfeeding and did not mention the potential for negative speech outcomes if they declined the surgery. This may be attributed to the paucity of clinical information available in the medical literature regarding the relationship between speech outcomes and frenotomy.

To the best of our knowledge, there are only four studies discussing the effects of frenotomy on speech outcomes [10–13]. Messner and Lalakea conducted a prospective study of 30 patients with ankyloglossia undergoing frenotomy, and were able to show significant objective improvement in tongue mobility, protrusion, elevation and speech outcomes [10]. In contrast, Dollberg et al. evaluated speech outcomes in eight pediatric patients (age 4–8 years) who underwent frenotomy and compared them to untreated and non-ankyloglossia patients. Although the treated children had fewer articulation problems than untreated children they did not find any difference in word, sentence or fluent speech intelligibility and concluded there was no significant difference in speech outcomes after frenotomy [12]. All of these studies are limited by small numbers, lack of control group or both and show that there is still controversy regarding the effect of frenulum reduction on speech.

Our study demonstrates that neonates who underwent frenotomy for breastfeeding difficulties maintained improved speech outcomes when compared to individuals that declined Surgical Intervention. Parents suggested a statistically significant improvement in three-year speech outcomes for those patients who underwent frenotomy when compared to untreated patients. In addition, there was no statistical difference in speech outcomes between the treated patients and normal controls.

In order to further assess the effect of frenotomy, parents were asked to describe the efficiency of their child's oral-motor activity. It is well understood that speech and oral motor function is strongly associated with the facial bone structure and mobility of muscles within the oral cavity such as the tongue [18,19]. We found that those patients treated with a frenotomy maintained an improved ability to use their tongue to clean the inside of their mouths after eating, clean the outside of their lips with their tongue and lick ice cream when compared to those who were untreated and were actually quite similar to the non-ankyloglossia control group. These findings are in accordance with previous studies conducted by Lalakea and Messner, in which patients saw improvements in the same oral motor activities [11].

Limitations of this study include those inherent to a retrospective survey. First, the survey with the Likert Scale is a subjective method of analysis and does not provide quantitative evaluation of vocabulary size or number of disarticulations, which is important to determine the severity of speech disorders. In addition, retrospective studies are inevitably susceptible to recall and selection bias. The parents may perceive a false positive effect in the child after a frenotomy procedure and vice versa. In an attempt to offset this potential bias, we utilized the Control Group during speech outcome analysis because these patients were not advised to obtain a frenotomy at any point during childhood. While it is difficult to reduce the described parental bias, this Control Group is important because it provides a cohort of patients outside of those who were evaluated by a lactation consultant or otolaryngologist at birth. Thus, we then compared their speech outcomes to the Surgical Intervention and No Surgical Intervention Groups, which

were advised to receive intervention. Because the Control Group patients were not advised to obtain frenotomy, we utilized their speech outcomes during our statistical evaluation to determine if there is significant speech improvement. Furthermore, a speech pathologist did not evaluate the child's speech outcomes when the telephone survey was conducted.

5. Conclusion

Ultimately, a long term randomized controlled trial to evaluate speech outcomes for those patients with ankyloglossia comparing those treated with frenotomy and those without would be ideal. Until that time, this study is the largest to date, with 71 patients in the treatment group, demonstrating improved speech outcomes in infants who underwent frenotomy for ankyloglossia.

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