



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

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