

Impact of ankyloglossia on the language development of children

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ABSTRACT

Objective: Ankyloglossia is a benign anomaly of the tongue which may cause functional limitation. Evidence regarding the impact of ankyloglossia on children's language development is limited. We aimed to evaluate the language development of children born with ankyloglossia.

Patients and Methods: Children diagnosed with ankyloglossia were followed up prospectively. Demographic characteristics, degree of ankyloglossia assessed by Hazelbaker score in infancy and its effects on breastfeeding were evaluated. Language development was tested by the Turkish version of the Test of Early Language Development-Third Edition and the Denver II Test at 3-5 years of age.

Results: Out of 53 children diagnosed with ankyloglossia, 38 (71.7%) children had language development testing and were included into the study. Significant ankyloglossia was detected in infancy in 10 of these children (26.3%). Median time of exclusively breastfeeding was not different according to the severity of ankyloglossia. All children evaluated with Denver II Test were developmentally normal in all domains. Scores of Test of Early Language Development-Third Edition were not different between children with and without significant ankyloglossia.

Conclusion: Long term language development of children with ankyloglossia was not adversely affected. Parents should be appropriately informed and efforts must be paid to prevent unnecessary surgical interventions concerning language delay.

Keywords: Ankyloglossia, Children, Language development

1. INTRODUCTION

Ankyloglossia, or tongue-tie, is a congenital anomaly of the tongue characterized by short, tight and thick frenulum [1]. The prevalence of ankyloglossia ranges from 0.1% to 10.7% in different populations with a male predominance. This range, in the reported prevalence is large because of the lack of a uniform definition and diagnostic criteria [2].

Ankyloglossia is usually a benign condition, which generally needs no intervention. However, if the mobility of the tongue is severely affected, it may result in functional limitations. These include breastfeeding difficulties in infancy causing significant nipple pain and nipple trauma in mothers. Traumatized nipples can cause the baby to remove milk less efficiently than baby without tongue-tie, therefore, the mother may have severe nipple pain due to incomplete breast drainage. In rare instances, interference with breastfeeding may be so severe that it may be the only cause of failure to

thrive [3]. The historical practice of treating ankyloglossia in all infants has already been dropped but there is still no consensus on how to approach ankyloglossia among medical specialists, such as pediatricians, plastic surgeons, otolaryngologists, lactation consultants and speech language pathologists [4]. Available studies about the effects of ankyloglossia on breastfeeding and its surgical management are heterogenous in terms of study design and definition of ankyloglossia [2,5,6]. It is not possible to establish a practice guideline for the management of ankyloglossia due to the low strength of available studies and the lack of knowledge on the natural course of ankyloglossia.

Even though, most of the studies on ankyloglossia are about the effects of breastfeeding, another concern related to ankyloglossia is speech outcomes in children. Available

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studies have conflicting results and the outcome measures are different [7-10]. Therefore, there is no universal consensus on how to approach ankyloglossia regarding future speech problems. Besides, studies on the speech outcomes of ankyloglossia focus on articulation disorders and social consequences but many parents also worry about their child's language development. In this study, our aim was to evaluate the language development of children born with ankyloglossia using objective tests and comparing children's language development according to the severity of ankyloglossia assessed in infancy.

2. PATIENTS and METHODS

Study design and participants

Children born with ankyloglossia between the years 2012 and 2015 and followed up at a well – child clinic at a university hospital were recruited into the study. Between the years 2012 and 2015, the clinic provided care for 2714 children and among them 53 children with ankyloglossia were identified during infancy and noted on personalized patient files. In the outpatient clinic, babies were regularly assessed according to the recommendations of the American Academy of Pediatrics. Physical examination, growth and developmental surveillance, immunization and anticipatory guidance were provided during the visits. For the aims of this study, patients' data regarding demographic characteristics, duration of breastfeeding and surgery for ankyloglossia were subtracted from each patient's personalized file. The degree of ankyloglossia was assessed in infancy by the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) in well – child visits by the pediatricians. HATLFF has five appearance items and seven function items [11]. Maximum score of appearance items is 10 and of function items is 14. Greater scores mean less severe ankyloglossia. Significant ankyloglossia was defined when the function score was less than 11 or the function score was 11 but appearance score was less than 10 or appearance item score was less than 8. Tongue-tie of 10 children (26.3%) was classified as significant during infancy. All children with significant ankyloglossia were referred to the plastic surgeon for further evaluation. The primary outcome of the study was the language development of the children. Duration of exclusive breastfeeding and total duration of breastfeeding were secondary outcomes. The study was approved by the Marmara University, School of Medicine Ethics Committee (Date: 05.10.2018, Number: 09.2018.674). This study was conducted in accordance with the Declaration of Helsinki.

Evaluation of global development and language

Families were called back at 3-5 years of age to assess the children's language development. Denver II Test was performed to examine the global development of children [12]. Language development was further evaluated specifically with the Turkish version of the Test of Early Language Development-3 (TELD-3)

[13,14]. A child developmental specialist who had certificates for Denver II and TELD-3 performed both tests. Written and verbal informed consent was obtained from parents. Since, the evaluation of articulation was beyond the scope of this study, no articulation tests were performed.

Denver II Test

All children were tested by Denver II for assessment of global development. Denver II is a screening test for the development of children aged 0-6 years and covers four developmental domains for testing: Fine motor, gross motor, personal-social and language skills. The Turkish version was validated in 1995 by Anlar and Yalaz [12]. Performance of children in Denver II was evaluated according to the children's age. Results were recorded in each domain according to child's performance as normal, suspicious or abnormal.

Test of Early Language Development-3

The Turkish version of the TELD-3 which was originally created by Hresko et al., was adapted by Topbaş and Güven in 2011 [13,14]. Both expressive and receptive language are assessed by TELD-3 and can be used for children between 2-7 months old [11]. The test consists of 2 forms; form A and form B, each with a 37-item receptive language subtest and a 39-item expressive language subtest. Scores were calculated first as raw scores and then converted into standard scores. In addition, we calculated the children's scores as age in months.

Statistical Analysis

Data analysis was performed by IBM SPSS statistical software (Version 20.0. Armonk, NY: IBM Corp.). Values for continuous variables were expressed as medians, interquartile range (IQR), means, standard deviations, minimum and maximum and compared by Mann-Whitney *U* test, after testing normal distribution by Shapiro-Wilk test. Descriptive variables were reported as numbers and percentages. Comparisons between categorical variables were performed by Chi-Square testing. *P* values of <0.05 were described as significant.

3. RESULTS

Fifty-three children were born with ankyloglossia between the years 2012-2015 and were followed up in our well – child outpatient clinic. In our cohort, the prevalence of ankyloglossia was identified as 2.0%. Out of 53 children, the families of 40 children accepted their child to be evaluated by Denver II and Turkish TELD-3. One boy in a bilingual family and another boy with a diagnosis of epilepsy were excluded from the study. 38 (71.7%) children had language development testing and were included into the study. All of the analyses described below were performed only on 38 children who were assessed for language development. Degree of ankyloglossia was assessed by HATLFF in infancy at a

median age of 2 (IQR:1-5) months. Significant ankyloglossia was detected in 10 (26.3%) children.

Most children were of male gender (n:25, 65.8%). The median age at the evaluation of language development was 56 months (IQR:51-60) and was not different between children with significant and non-significant ankyloglossia. Four children (10.5%) were the product of consanguineous marriage and 10 (26.3%) children had a family history positive for ankyloglossia (Table I).

Table I. Comparison of children with significant and non-significant tongue-tie

	Significant	Non-significant	P value	Total
Number of children, n (%)	10 (26.3)	28 (73.7)	NA	38 (100)
Age, months, median (IQR)	58.5 (53.2-62.5)	56.0 (49.5-59.0)	NS	56 (51-60)
Gender, male, n (%)	6 (24.0)	19 (76.0)	NS	25 (100)
Consanguinity, n (%)	2 (50.0)	2 (50.0)	NS	4 (100.0)
Family history of ankyloglossia, n (%)	4 (40.0)	6 (60.0)	NS	10 (100.0)
Exclusive breastfeeding, months, median (IQR)	6.0 (0.7-6.0)	6.0 (5.0-6.0)	NS	6.0 (5.0-6.0)
Total breastfeeding, months, median (IQR)	21.0 (10.7-25.5)	24.0 (11.7-24.0)	NS	24.0 (12.0-24.0)
Denver-II, Normal, n (%)	10 (26.3)	28 (73.7)	NS	38 (100)

IQR: interquartile range, NA: Not applicable, NS: Non-significant

Most of the children (n:28, 71.8%) were exclusively breastfed in the first 6 months. Median duration of exclusive breastfeeding was 6 (IQR:5-6) months and total breastfeeding was 24 (IQR:12-24) months. Duration of exclusive breastfeeding and total breastfeeding were not different between children with significant and non-significant ankyloglossia (Table I). None of the infants required further breastfeeding counseling other than that which was provided during regular well-child visits.

All children with significant ankyloglossia according to HATLFF were also examined by a plastic surgeon. Eight children (21.1%) were operated at a median age of 7 (IQR: 3.2-21) months. Surgery was offered to all children with significant ankyloglossia according to the consensus decision of the pediatricians and the plastic surgeon and was performed in three children with significant ankyloglossia while the families of seven children with significant ankyloglossia opted not to be operated. Additionally, the families of five children with non-significant ankyloglossia had their children's tongue-tie operated elsewhere, even though, surgery was not recommended by our team. A history of the operation was gained through interviewing the parents and was confirmed by physical check. There were no significant differences in HATLFF appearance and function scores between the children who were operated and those who were not. Duration of exclusive breastfeeding and total breastfeeding did not show any differences between children who had undergone tongue-tie division and those who had not.

All children were developmentally normal. The results of Denver II and TELD-3 expressed as age in months of all children were shown in Tables I and II. All children were evaluated as normal in all domains of Denver II. Accordingly, none of the families described any speech delay. The results of TELD-3 were not statistically different in children with and without significant ankyloglossia according to HATLFF score. There were also no differences of TELD-3 scores in children who had frenotomy (n=8) and those who did not (n=30).

Table II. TELD-3 results of children with significant and non-significant ankyloglossia. No significant differences were detected between the two groups in TELD-3 receptive language scores (P=0.33) and expressive language scores (P=0.25).

	Age		TELD-3 Receptive Language		TELD-3 Expressive Language	
	Significant	Nonsignificant	Significant	Nonsignificant	Significant	Nonsignificant
	n=10	n=28	n=10	n=28	n=10	n=28
Mean ±SD	57.2±7.4	54.6±7.6	60.1±9.4	58.4±8.8	59.9±9.3	58.4±9.2
Median (IQR)	58.5 (53.2-62.5)	56 (49.5-59)	62 (58.7-64)	61 (52-63)	62.5 (58.5-63.2)	61 (49.5-63)
Minimum	40	40	38	40	38	40
Maximum	66	77	74	85	73	85

IQR: interquartile range, SD: standard deviation, TELD-3: Test of Early Language Development-3.

All values are represented in months

4. DISCUSSION

Most of the studies regarding ankyloglossia focused on breastfeeding issues. However, in daily clinical practice, parents often share with physicians their concerns about the late outcomes of ankyloglossia. Speech problems are one of the major concerns of parents but there is inadequate evidence to objectively inform parents about the effects of ankyloglossia on the language development. In our study, we performed specific language development tests in addition to evaluation of global development and showed that children with ankyloglossia were able to present age – appropriate language development skills regardless of the severity of their tongue-tie assessed during infancy, whether operated or not.

Clinical significance of ankyloglossia has been a subject of debate among medical professionals. In a survey conducted among otolaryngologists, pediatricians, lactation consultants and speech pathologists, it has been shown that there is no consensus about the significance of ankyloglossia regarding speech, breastfeeding and surgery outcomes [4]. Some of the authors propose that neonatal frenulum is naturally short and with age and repeated use, the short frenulum may elongate and will not necessarily result in functional problems [15-17]. The lower incidence in adults may support this idea [1,2]. However, no study prospectively evaluated the natural course of ankyloglossia to confirm this assumption. On the contrary, there is a body of evidence that it may cause infant feeding difficulties, speech and articulation disorders and social distress [1,18,19]. Some parents may also think that ankyloglossia may be a reason for speech delay, but children with ankyloglossia are expected to follow regular developmental milestones in language, as we showed in our study, although, to our knowledge no prior study has investigated the language development in children with ankyloglossia.

Speech problems are the second most prevalent outcome described in the ankyloglossia literature [7]. Concerns related to speech is one of the main reasons why parents seek for frenotomy [20,21]. However, studies are insufficient to prove a causative association of ankyloglossia with speech disorders [9]. There is some literature reporting an improvement in speech through treatment, but results commonly rely on parent reporting without standardized measurement tools for speech outcomes [8,21]. One of the distinguishing features of our study from other reports in the literature is that we performed standardized tests to assess the language development of the children. In the study of Klockars et al., the most common indication for tongue-tie division was speech and articulation problems and 84% of parents reported a benefit from the surgery, but in this study, it was not clear how speech problems were evaluated and diagnosed. In addition, the benefit of the operation was reported only by parents without any objective measurements [21]. Walls et al., questioned parents regarding the speech intelligibility of their children three years after neonatal frenotomy and compared results with a control group and with a non-surgical intervention group. Parental subjective evaluation of speech regarding vocabulary, articulation and sentence structure were graded higher by a Likert Scale survey in the group of surgically treated children [8]. In contrast, Daggumati et al., found no parenteral perceived difference in speech quality between surgically treated and conservatively

managed children [22]. In four studies, speech was evaluated by speech-language pathologists rather than objective parental assessment. Heller et al., compared two surgical methods in 16 patients with persistent articulation problems and presented that 4-flap Z-frenuloplasty provided better improvement in articulation judged by speech-language pathologists [23]. The test used by speech language pathologists was not identified in this study. Similarly, in the study of Messner et al., 82% patients with ankyloglossia were reported to have improved speech postoperatively judged by nonblind speech-language pathologists without a standardized test [18]. In two studies, standardized articulation tests were used. Ito et al., from Japan evaluated changes in speech by a standardized articulation test in five patients. They concluded that articulation errors can be improved after tongue-tie division, even though their study was limited by a low number of patients, nonblind assessment of speech and lack of a control group [24]. In the retrospective cohort of Dollberg et al., speech intelligibility of children with untreated and treated ankyloglossia with an age matched control group was assessed by a standardized articulation test. Articulation errors were more common in children with untreated ankyloglossia but there was no statistical difference in word, sentence and fluent-speech intelligibility [25]. Recently, Salt et al., revealed that objective measures of speech outcomes did not differ between treated-, untreated-, and non-tongue tie groups [26]. These studies with conflicting results cause confusion and discrepancy among medical specialists who encounter ankyloglossia. Moreover, studies investigating the effects of ankyloglossia on speech are focused on articulation rather than language development which is often a point of discussion with parents of children with ankyloglossia in well child visits. At this point, we believe that our study adds to the limited evidence about language development in children with ankyloglossia since we used language development tools in the assessment. It has been shown that parents are the main decision makers for frenotomy following lactation consultants [20]. Therefore, adequate and reliable information must be given to families by health care professionals caring for newborns and infants. We believe that our study provides evidence to reassure parents that children with ankyloglossia exhibit normal language development same as their peers. Therefore, delayed language development in children with ankyloglossia should never be solely attributed to ankyloglossia and a usual evaluation including neurodevelopmental and audiologic assessment should not be deferred while waiting for the benefits of surgery.

Ankyloglossia has been blamed for interfering with successful breastfeeding. As part of efforts to increase breastfeeding rates, there is a substantial increase in the diagnosis and surgical treatment of ankyloglossia [27-29]. However, studies are heterogeneously designed and not enough to recommend frenotomy to improve breastfeeding [6]. In the recent Cochrane review, it has been emphasized that frenotomy reduces maternal nipple pain in the short term evaluation but does not have a consistent positive effect on breastfeeding [30]. Furthermore, no study investigated the effects of ankyloglossia on long-term breastfeeding and resolution of maternal pain whether frenotomy is performed or not. Available studies did not study whether

infants were breastfed for longer after frenotomy [6,30]. In our study, we were able to show that the mean durations of exclusive and total breastfeeding were not different between infants who had frenotomy and who those had not. Besides, breastfeeding duration also did not change according to the severity of ankyloglossia assessed by HATLFF. In our well-child clinic, mothers are informed about the importance of breastfeeding and counselled on successful breastfeeding. Breastfeeding issues are discussed in every visit from the first visit on and lactation specialists observe mother-infant dyad for breastfeeding at least once and every time the mothers have a concern. According to World Health Organization (WHO), infants should be exclusively breastfed for the first six months and continue breastfeeding up to the age of two years or beyond [31]. In our study group, infants were exclusively breastfed for a median of 6 months and median duration of total breastfeeding was 24 months consistent with the WHO recommendations. Breastfeeding counselling efforts in our clinic may have resulted in the success of mothers with tongue-tied infants and may have prevented unnecessary interventions. Therefore, intensive lactation support should be implemented before tongue-tie is blamed for the failure of breastfeeding. In line with this, Dixon et al., noticed that breastfeeding rates remained stable despite an increase in frenotomy rates in the last years and showed in their study that a multidisciplinary program including objective assessment of tongue function and anatomy along with lactation support is effective in reducing frenotomy rates while improving breastfeeding [32]. Muldoon et al., revealed that frenotomy reduced self-reported maternal nipple pain and provided an overall improvement in breastfeeding but rates of exclusive breastfeeding remained unchanged following frenotomy compared with pre-frenotomy [20]. These results show that frenotomy does not always resolve breastfeeding difficulties and highlight the importance of breastfeeding counselling for mother-infant dyads by experienced health care professionals.

Our study has several limitations. Our sample size may seem too small to generalize our findings for all patients with ankyloglossia but we prospectively followed all patients with ankyloglossia in our cohort. Besides, the prevalence of ankyloglossia in our cohort (2%) was within the range of the reported prevalence in the literature [2]. Another limitation is that the natural course in patients who had undergone frenotomy according to the family request is unknown. Even though, articulation was also evaluated while performing language development tests to inform the parents, a standardized articulation test was not used, since it was not the scope of this study. Other limitations are that there was no control group to compare the language development of children with ankyloglossia and the groups which were compared were uneven in number. Despite these limitations, we believe that our study adds to the limited literature comparing speech outcomes of ankyloglossia between treatment and nontreatment groups, as most of the studies compared surgical modalities or changes before and after surgical intervention. Due to prospective design, we were also able to watch the natural course of ankyloglossia in patients who had no frenotomy. Besides, to our knowledge, this is the first study evaluating objectively the language development of children with ankyloglossia. Furthermore, even though there is

no universally accepted method of scoring for ankyloglossia, our study is one of the rare studies in which patients were assessed objectively by a validated tool in infancy. The decision on surgery was not only based on scoring but also on consensus reached between pediatric and plastic surgery teams, except for the five patients who were operated upon family request.

In conclusion, language development of children born with ankyloglossia are expected to follow regular steps as normal children. Families with a concern of speech delay should be informed comprehensively to avoid unnecessary surgical interventions which may prevent timely evaluation and management of language delay. Evidence-based guidance on treatment of ankyloglossia is still lacking. Communication between pediatric teams and surgeons is essential in determining the need for surgery until evidence-based guidelines are constituted. Future prospective studies with a larger sample size and long term follow up in which tongue – tie severity is assessed relying on objective tests, would be ideal for a comparison between patients who were surgically treated and those who were not.

Compliance with Ethical Standards

Ethical Approval: The study was approved by the Marmara University, School of Medicine Ethics Committee (Date: 05.10.2018, Number: 09.2018.674). This study was conducted in accordance with the Declaration of Helsinki. Written and verbal informed consent was obtained from parents.

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