



Prevalence of tongue-tie and evaluation of speech sound disorder in young children

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Purpose: Tongue-tie is a common congenital abnormality whose effect on speech sound disorders is controversial. The first purpose of this study was to determine the prevalence of tongue-tie in Persian speaking children aged 3–6 years. Second, compare the speech sound errors of children with tongue-tie with those of children without tongue-tie.

Methods: 487 (206 female, 281 male) children aged 3–6 years were recruited for this study. Descriptive statistics were used to determine the prevalence of tongue-tie. Fisher's exact test was performed to compare the speech sound errors of children with and without tongue-tie.

Results: The prevalence of tongue-tie in children was 14.9%. These children showed a statistically significant difference in the production of the /s/, /z/, and /l/ sounds in both the initial and final positions of the word and the /t/ sound in the initial position of the word.

Conclusions: While it is theoretically expected that the tongue-tie affects the production of anterior sounds, our clinical study shows speech sound disorder in only a limited number of cases. Therefore, it seems that surgical intervention on tongue-tie for sole purpose of speech improvement should be performed cautiously.

Keywords: Tongue-tie, Ankyloglossia, Speech sound disorder, Prevalence

INTRODUCTION

Ankyloglossia, commonly referred to tongue-tie, is a congenital anomaly characterized by an abnormally short lingual frenulum [1-4]. The prevalence of tongue-tie has been reported to range from 2.33% to 32.54% [5-8] with a higher prevalence in males than females with a ratio of 3.1 [2,9-12].

Several studies have reported limited function and movement of tongue as a consequence of tongue-tie [2,3,11,13,14]. Tongue-tie may cause problems such as sucking and swallowing difficulties as well as weight gain and mouth breathing in infancy [1, 15]. In addition, tongue movements play an important role in speech production, and therefore the tongue-tie can negatively affect speech production, resulting in speech sound disorder (SSD). Messner and Lalakea reported 71% of young children with tongue-tie had speech problems related to restricted tongue mobility [14]. Klockars and Pikaranta, conducted a study using a detailed patient questionnaire and reported that the main indication for tongue-tie division is articulation problems (64%) [10]. Us-



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ing Van der Merwe 's model of sensorimotor speech control [16], the possible effects of tongue-tie can be interpreted. According to this model, four levels of neurological structure and phases of speech processing are considered, which include linguistic symbolic planning, speech motor planning, speech motor programming, and execution. Restrictions in the movement of the tongue as a result of tongue-tie may negatively affect the execution phase and the production of speech oral sounds.

Walls and colleagues compared three years old children in three groups; surgical treated children for tongue-tie (n=71), children with tongue-tie (n=15), and control group (n=18). This survey was conducted using a Likert scale to assess speech perception by parents. The results of the study displayed that the speech in the first group was better than the second group, and in the second group was better than the last group [17]. Additionally, Messner and Lalakea studied children with ankyloglossia aged 1 to 12 years undergoing frenuloplasty. Twenty-one of children who were aged 2 or older underwent formal speech evaluations preoperatively and 15 (71%) of 21 children had articulation disorder. Postoperative evaluation in 15 of these children showed improvement in consonant articulation in 9 children. In this study speech sound disorder reported by yes or no question [14]. These researchers conducted another study on fifteen individuals with tongue-tie ranging in age from 14 to 68 years. Participants completed a written questionnaire regarding possible speech effects of ankyloglossia. The most common single problem noted by participants was speech difficulty (50%) compared to control subjects without ankyloglossia [13]. Ito and Shimizu et al. [18] designed a study to determine the effectiveness of tongue-tie division for speech articulation disorder in five 3-8 years old children with tongue-tie. The speech was evaluated by 50 pictures of common Japanese words with 2-3 syllables. The patients primarily had substitution, omission and misarticulation of the consonants such as *dʒ*, *ts*, *ʃ*, *r*, *s*, *t*, *d*, *dz*, and *ts*. Substitution and omission improved relatively early after tongue-tie division and progressed to distortion, which is a less-impaired form of articulation disorder [18]. In this study standardized articulation tests were used in order to assess speech disorder. In another study Functional Improvements of speech after lingual Frenectomy Tongue-Tie release was evaluated using a questionnaire which is filled by parents. Speech improved in 89% of Thirty-seven participants aged 13 months to 12 years [19].

However, Salt et al. [20] studied three groups of children

aged 2; 1 to 4; 11 years, including children in whom the tongue was divided because of a tongue-tie (n=21), children with a tongue tie (n=17), and children without a tongue tie (n=21), to understand the relationship between tongue-tie and speech production. In this study, no significant difference in speech production was found between children with and without tongue-tie [20]. It is worth to mention that in this study 4 children of treated group had speech therapy program for /l/,/s/,/r/,/ʃ/,/tw/,/θ/,/z/,/f/, and 2 children in untreated group for /s/.

In another study, Dollberg et al. [21] compared consonant production of children with treated tongue-tie (n=8) and children without tongue-tie (n=8) as well as children with untreated tongue-tie (n=7). All the children were 4-8 years old. The articulation of /t/,/d/,/l/,/r/ consonants were not significantly different between children with treated and untreated tongue-tie. Moreover, there was no significant difference between treated children and children without tongue-tie [21].

While several studies have evaluated the association between tongue tie and oral speech sound errors, the directionality of this relationship is still unclear.

Although several studies have examined the association between tongue-tie and the production of oral sounds, inconclusive results have been reported due to small sample size, different tongue-tie assessment instruments and history of speech therapy in children with tongue-tie. Also, a survey showed that the beliefs of experts regarding the tongue-tie and speech problems are different. The results of that study revealed sixty percent of Otolaryngologists, 50% of speech pathologists, and 23% of pediatricians believe tongue-tie is at least sometimes associated with speech difficulties [4]. In addition, to the best of our knowledge just two studies of Ito et al. [18] and Dollberg et al. [21] were conducted in non-English language [18,21] which may limit the generalizability of the results. So, as highlighted in a systematic review by Wang et al. [22], there is a need for further research in this area.

The first aim of the present study was to determine the prevalence of tongue-tie in three- to six-year-old Persian-speaking children. Due to the conflicting results on the effect of tongue tie on oral speech production, the second aim of this study was to evaluate the speech sounds of children with tongue-tie using a standardized speech sound disorder test [23] and compare them with those without this complication.

METHODS

Sample study

The participants of this study included 487 healthy (206 females, 281 males) children between three to six years old (Table 1). They were randomly selected from kindergarten children via cluster sampling and were assigned to two groups based on the presence of tongue-tie. No history of speech therapy, orofacial anomalies, motor speech disorders, and speech-language developmental delay have been reported for participants in this study. First, all participants were assessed using assessment score for ankyloglossia in children to evaluate the presence of tongue tie (Table 2). According to Ito et al. [18] tongue-tie is considered to be diagnosed where the total achieved score is less than 8 out of 10 total which represents a normal case. This assessment was done by three speech-language pathologists with three years of experience in speech sounds disorders and intraoral examination.

After diagnosing children with tongue-tie, the single word quick repetition test was used to evaluate the speech sound disorders between two groups of children with and without tongue-tie. Single word quick repetition test is an assessment tool used to evaluate the speech sound abilities of Persian speaking children aged 3 to 8 years [24]. This test included 30 words with CVC structure and the maximum total score for the test is 44 target phonemes. This study was reviewed and approved by the Ethics Committee of Mashhad University of Medical Sciences [IR.MUMS.REC.1398.071].

Inter-rater agreement

In order to estimate the inter-rater agreement of our tests, 10 children were evaluated on the same day, simultaneously and independently by three speech language pathologists.

Data analysis

All analyses were conducted with SPSS Version 19 (IBM,

2010). Descriptive statistics were used to determine the prevalence of tongue tie and correct percent for each target consonant production for each age group. Fisher’s exact test was performed to compare the correct speech sound production between the two groups of children with and without tongue-tie. Nevertheless, to evaluate the level of agreement among the three evaluators, the percent agreement was calculated.

RESULTS

Among 483 children (206 females and 281 males) aged 3-6 years (mean ± SD = 57.69 ± 10.08 months), 72 were children (24 females and 48 males) with tongue-tie. Age of children with tongue-tie (mean ± SD = 56.39 ± 8.78) were not statistically different from the age of children without tongue-tie (mean ± SD = 57.92 ± 10.27) (*p* value = 0.189). Percent agreement among evaluators for assessment score for ankyloglossia was 90% and for evaluating the speech sound disorders was 100%.

The highest percentages of tongue tie (27.8%) were reported in children between 4;6 to 4;11 years old. The lowest percent-

Table 2. Assessment score for ankyloglossia in children (Ito et al., 2015)

Attachment of lingual frenulum to tongue	2: >1 cm from tip 1: >6 mm and <1 cm from tip 0: <5 mm from tip
Attachment of lingual frenulum to oral floor	2: Attached to floor of mouth 1: Attached to lower ridge 0: Attached to upper ridge
Appearance of tongue when protruded	2: Round 1: Slight cleft in tip 0: Heart-shaped
Extension of tongue	2: Tip over lower lip 1: Tip over lower gum 0: Neither of the above
Speech for age	2: not disturbed 1: a little disturbed 0: markedly disturbed

Table 1. Participant characteristics for the children with and without tongue-tie

		3;0-3;5	3;6-3;11	4-4;5	4;6- 4;11	5- 5;5	5;6-5;11	Total
Number of Children without tongue-tie	Male	19	39	28	40	38	68	232
	Female	15	23	30	40	30	41	179
	Total	34 (8.3%)	62 (15.1%)	58 (14.1%)	80 (19.5%)	68 (16.5%)	109 (26.5%)	411 (100%)
Number of Children with tongue-tie	Male	3	5	10	17	5	8	48
	Female	2	2	8	3	6	3	24
	Total	5 (6.9%)	7 (9.7%)	18 (25%)	20 (27.8%)	11 (15.3%)	11 (15.3%)	72 (100%)

ages of correct speech sounds were found in /ʒ/ in both groups of children with and without tongue tie (Table 3). In addition, among children with and without tongue tie, the highest percentages of correct speech sound were /n/ and /l/, respectively (Table 3). All sounds, except /ʒ/, correctly produced by more than 96% of the children without tongue-tie. In contrast, /ʒ/, /s/, and /z/ were correctly produced by less than 90% of children with that disorder.

The Fisher's exact test revealed significant differences in correct articulated consonants between two groups of children with and without tongue tie for /s/, /z/, and /l/ in both initial (p value=0.000, p value=0.000, and p value=0.031 respectively) and final position (p value=0.000, p value=0.000 and p value=0.001 respectively). In addition, the sound /t/ in the initial position showed significant difference between two groups of children (p value=0.001).

DISCUSSION

The aim of this study was to determine the prevalence of

tongue-tie in three to six years-old Persian-speaking children and to compare the correct speech sounds in children with and without tongue-tie. The results of the current study showed that out of 483 children, 14.9% ($n=72$) had the short lingual frenulum. The prevalence rate of this complication varies from 4% to 32.5% in different studies [3,6,8], and the percentages obtained in the present study fell within the aforementioned range. This discrepancy between the different studies may be attributed to variations in the age of the subjects, the assessment and diagnostic criteria, as well as the number of participants.

In addition, there were significant differences between children with and without tongue tie for /s/, /z/, and /l/ in both initial and final position and /t/ in initial position. Errors for /s/ and /z/ consonants were distortion and for /t/ was omission. /l/ sound error was omission in some children and substitution by /j/ sound in others. The speech errors in children with tongue-tie usually can be attributed to the structural defect and limited motor range and function of tongue and considered as phonetic error.

While previous studies have reported differences between children with and without tongue-tie in the production of sounds, this study reported percent correct consonants in both groups and then evaluated the differences between them. The results of the present study show that some children in both groups had problems with the production of anterior sounds. However, there were significant differences between children with and without tongue tie for /s/, /z/, and /l/ in both initial and final position and for the sounds /t/ in initial position which is consistent with some previous studies [10,11,13,14,18]. For example, in a study of Japanese language, the /t/, /d/, /n/, /s/, /z/, /r/ sounds were impaired in children with tongue-tie. It should be noted that /th/ and /l/ sounds do not occur in Japanese; therefore, they were not evaluated in this language [18].

In the current study, it seems that problems in the articulation of sounds are not due to the age of consonant acquisition in children: among the consonants whose mastery ages are high, there exists consonants where the results did not show any statistical differences between two groups; for example, /ʒ/ which its mastery age in the Persian language for initial position is up to 5 years and for final position is up to 6 [23], while for some other consonants with the same age of mastery in articulation, like /z/, results showed a significantly different between two groups. Similarly, for the consonants with a low age of mastery, in some cases the results revealed a dif-

Table 3. Percent of correct consonants in each group of participants

Consonants	Children with tongue-tie	Children without tongue-tie
N _i *	100	99.5
N _r **	95.8	98.5
L _i	93.1	98.1
L _r	91.7	99
S _i	83.3	98.8
S _r	80.6	99.8
ʃ _i	93.1	96.1
ʃ _r	90.3	97.1
ʒ _i	69.4	67.7
ʒ _r	45.8	58.2
Z _i	77.8	97.3
Z _r	83.3	97.8
t _i	93.1	99.5
t _r	97.2	99.3
d _i	100	99.5
d _r	95.8	97.8
tʃ _i	95.8	95.6
tʃ _r	94.4	97.1
dʒ _i	95.8	96.8
dʒ _r	97.2	93.9

i*: initial position of word; r**: final position of word.

ference between the two groups, for instance, in the case of /t/ with the mastery age of 3 in Persian language [23]. Whereas /n/ with the same age of mastery had no significant difference between children with and without tongue-tie. Considering all these, still it is suggested to interpret the data with caution.

In contrast with our results, one study which was performed on Hebrew language showed children with treated tongue-tie have not any significant difference with children without tongue-tie in sound production. In another part of this study, results indicated that although untreated children have problems in production of /t/, /d/, /l/, and /r/, this difference was not statistically significant [21]. Small sample size of each group may affect the results of this study. Salt et al., also, reported that children with tongue-tie did not differ in sound production from children without this disorder and children with treated tongue-tie [20]. Nevertheless, in the mentioned study, 19% of children with tongue-tie had a history of speech therapy in order to correct sounds production (e.g., /l/, /s/, /r/, /ʃ/, and /z/).

All studies did not report the same disordered sounds in children with short lingual frenulum although some of them were common. Different disordered sounds in these children can suggest that some of these children may compensate for limited tongue mobility without repair or surgery [13]. Finally, the results of the present study and other similar studies [10, 11,13,14,18] suggest that tongue-tie can affect sound production for at least some anterior sounds in some children.

Limitations and future directions

The insufficient cooperation of three-year-old children was one of the limitations of this study and the reason for the lower number of children in this age group compared with other age groups. In addition, the present study did not distinguish between different types of tongue-tie (superior and inferior attachment). Therefore, one possible direction for future studies is to study higher number of subjects of both genders in each age group in order to compare the differences between them by gender and age. Additionally, another extension could be to consider the type and severity of the tongue-tie to determine its effects on speech sound disorders.

CONCLUSION

Although theoretically the tongue-tie might be expected to affect the production of anterior sounds, our clinical evaluations show speech sound disorder in only a limited number of

cases.

In the present study, 72 of 483 studied children (14.9%) were diagnosed with tongue-tie. It was found that these 72 children had speech sound disorder (SSD) in the production of the /s/, /z/, and /l/ sounds in initial and final positions, and the /t/ sound in initial position compared with children without this disorder. Therefore, it appears that surgical intervention on the lingual frenulum for the sole purpose of improving speech should be undertaken with caution. These results may also help to develop guidelines for the management of ankyloglossia.

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