



Speech production in young children with tongue-tie

Holly Salt^a, Mary Claessen^a, Timothy Johnston^b, Sharon Smart^{a,*}

^a Curtin University, Kent St, Bentley, WA, Australia, 6102

^b IKids Paediatric Dental Care, 94 Stirling Hwy, North Fremantle, WA, Australia, 6159



ARTICLE INFO

Keywords:

Tongue-tie
Ankyloglossia
Speech
Intelligibility
Tongue mobility
Children

ABSTRACT

Introduction: Tongue-tie, or ankyloglossia, occurs in 4–10% of the population. Treatment of tongue-tie has increased by 420% in Australia between 2006 and 2016 and 866% in the United States between 1997 and 2012. Despite limited evidence, it has been suggested that tongue-tie can result in speech sound disorder (SSD). This study aimed to investigate tongue mobility and speech production outcomes in children with and without tongue-tie diagnoses.

Method: Fifty-nine children aged 2; 1 to 4; 11 years were recruited and formed three groups: treated tongue-tie (TTT), untreated tongue-tie (UTT) and no tongue-tie (NTT). Measures of lingual frenulum structure and function, tongue mobility, speech production, and parent and clinician intelligibility ratings were collected.

Results: No statistically significant differences were found between the TTT, UTT and NTT groups for tongue mobility, speech production or intelligibility. Significantly more UTT children had a history of speech pathology attendance than participants in the NTT group.

Conclusion: This study provides preliminary evidence of no difference between tongue mobility and speech outcomes in young children with or without intervention for tongue-tie during infancy. This study assists with clinical decision making and makes recommendations for families not to proceed with surgical intervention for tongue-tie during infancy, for the sole outcome of improving speech production later in life.

1. Introduction

Tongue-tie, or ankyloglossia, has traditionally been described as a congenital oral abnormality where movement of the tongue is restricted by a shortened, tightened or thickened lingual frenulum that connects the tongue to the floor of the mouth [1]. Recently, Mills et al. [2] examined the anatomy and histology of cadavers and noted the lingual frenulum is a midline fold rather than a band as previously thought [2]. Recently, a panel of ankyloglossia experts in the United States collaborated to form a Tongue-Tie Case Definition (TTCD) for newborns from birth to six months. They proposed that tongue-tie is defined anatomically in addition to functional or behavioral impairments [3]. An observable or palpable lingual frenulum is considered normal and is common in young children, so should not be considered a tongue-tie in the absence of functional impact [4].

The prevalence of tongue-tie varies between 4 and 10% [5], with two studies from the United States reporting 4.2% and 4.24% prevalence using the Hazelbaker Assessment of Lingual Frenulum Function (HATLFF) [6,7]. Additional rates of 4.2% and 10.7% were reported using subjective observations in United States and the United Kingdom,

respectively [8,9]. In contrast, a much higher rate of 32.34% was reported in Brazil during visual inspection of posterior tongue-tie in infants [10]. Treatment of tongue-tie has increased worldwide. In the United States, there were significantly more cases diagnosed with ankyloglossia (834%) and also more frenulotomy procedures (866%) reported between 1997 and 2012 [11]. A similar 420% increase was found in Australia between 2006 and 2016 [12]. The recognition of infant and maternal health benefits associated with breastfeeding has contributed to increased global promotion of breastfeeding [13,14]. As the presence of a tongue-tie in an infant can exacerbate feeding difficulties, this has likely contributed to the increase in ankyloglossia procedures [15].

There are many methods to assess and classify tongue-tie based on appearance and function [16]. Hong et al. [17] propose classifying attachment of the frenulum as either anterior or posterior, while Genna and Coryllos [18] suggest a classification system that uses four categories. These range from a fine elastic frenulum with an associated heart-shaped tongue and attachment points of tongue-tip and near alveolar ridge, to a thick, non-visible frenulum from the base of the tongue to the floor of the mouth which can be palpated. Suter and

* Corresponding author. School of Occupational Therapy, Social Work and Speech Pathology, Curtin University, GPO Box U1987, Perth, WA, 6845, Australia.

E-mail addresses: holly.salt@graduate.curtin.edu.au (H. Salt), m.claessen@curtin.edu.au (M. Claessen), timjohnston@westnet.com.au (T. Johnston), Sharon.Smart@curtin.edu.au, sharon.smart@curtin.edu.au (S. Smart).

<https://doi.org/10.1016/j.ijporl.2020.110035>

Received 23 December 2019; Received in revised form 1 April 2020; Accepted 1 April 2020

Available online 04 April 2020

0165-5876/ © 2020 Elsevier B.V. All rights reserved.

Borstein [19] provide alternative methods of assessing tongue-tie structure including measuring length of free tongue, frenulum length and use of the Frenotomy Decision Rule for Breastfeeding Infants (FRDRI) [20].

There are a number of studies that suggest frenulotomy is beneficial for improving breastfeeding of infants with tongue-tie, by improving nipple pain, sucking and latch [15], though there is less evidence on the effects of tongue-tie on speech outcomes of children. Despite limited quality evidence to support claims, it is widely believed that tongue-tie can result in speech sound disorder (SSD) [15]. Bellinger et al. conducted a longitudinal cohort study of children who had tongue-tie treatment in the neonatal period, and documented parent reports for dental and speech outcomes at 9 and 38 month of age [21]. They found that only two out of 112 (1.8%) parents reported concerns with speech at 38 months of age. A systematic review investigated the impact of tongue-tie surgery on both speech and feeding, and found no causal relationship between tongue-tie and speech production [22]. Other studies have reported improvements in speech outcomes in individuals with tongue-tie who underwent surgery, but have used measures including 'subjective gains in speech', parent perception of speech intelligibility, presence of articulation errors and 'speech pathology evaluation'. These studies included small samples sizes and many documented improvements were not statistically significant [23–25]. A recent case series report from five participants found improved speech skills (as measured by parental reports, and increased accuracy in production of some speech sounds) following laser frenectomy. They also reported that the participants required ongoing speech pathology services [26]. Similarly, a case series reported the outcomes of five children and found less severe speech sound processes, including improvements in substitution and omissions following tongue-tie surgery [27]. Two studies have used parent intelligibility ratings to measure speech production and found ratings significantly improved following surgery [24,28]. Though parent intelligibility ratings are a clinically valid observation, both studies lacked objective use of speech measurement [24,28,29]. A Portuguese study discovered that children and adults who had frenectomy and speech pathology intervention exhibited improved tongue mobility, though their ability to produce the sound under investigation, a rhotic phoneme called the alveolar tap, did not improve [30]. A recent retrospective publication investigated the outcomes of 48 participants with tongue-tie. They found that children with moderate-to-severe, or severe speech or language difficulties experienced greater improvement of symptoms following frenulectomy than children with mild and mild to moderate difficulties [31]. Finally, a well-designed study by Dollberg and colleagues compared speech outcomes of 23 children aged four to eight years old [32]. They compared three groups: tongue-tie treated with frenulotomy conducted during infancy (between two days and four weeks), untreated tongue-tie and no history of tongue-tie. Articulation, speech intelligibility, oral anatomy and tongue movement assessments revealed no significant differences between groups on each measure. Given the small sample size within the study, the findings were interpreted with caution [32]. In conclusion, the current evidence highlights the need for further research in this area, with an adequate sample size, control group and objective in addition to subjective measures of speech production [33].

The potential effects of tongue-tie on speech production can be interpreted theoretically using the model of speech sensorimotor control by Van der Merwe [34]. This framework consists of neural structures and phases in processing of speech, through four levels: linguistic symbolic planning, speech motor planning, speech motor programming and execution. When an individual presents with tongue-tie, the movement of their tongue, a prime articulator in speech, can be restricted [1]. This suggests the 'impairment' of tongue-tie lies in the execution stage of the model. When this breakdown occurs, tactile-kinesthetic feedback is relayed back to the speech motor program, providing information on mobility parameters of the tongue such as muscle tone, velocity, range, direction and force. An alternative motor program

is created and executed accounting for the impairment as a result of tongue-tie. The output of this speech is therefore an altered version, creating the differentiation in speech production between some individuals with and without tongue-tie.

The purpose of this study was to explore the relationship between tongue-tie status and speech production with a larger sample size than previous studies, inclusion of a control group and use of both objective and subjective measures. The relationship between lingual frenulum function and speech production was investigated by comparing children with no tongue-tie diagnosis (NTT) to children with untreated tongue-tie (UTT), and the effectiveness of frenulotomy on speech production was explored, by comparing the NTT group to a group of treated tongue-tie (TTT) children. This pilot study aimed to investigate whether:

1. Tongue-tie significantly impacts speech production as measured by standardised speech production assessments.
2. Tongue-tie significantly impacts intelligibility (clinician and parent) as measured by a Likert scale intelligibility rating.
3. Tongue-tie significantly impacts tongue mobility as measured by the lingual control total score on the Verbal Motor Production Assessment for Children (VMPAC).

2. Methods

2.1. Participants

Fifty-nine children aged between 2; 11 and 4; 11 years were recruited to form three groups of participants: TTT, UTT and NTT. See Table 1 for characteristics of participants.

Participants in the TTT and UTT groups were recruited from iKinds Specialist Paediatric Dentistry in Perth, Western Australia and the NTT participants via convenience sampling. All children were required to have English as their primary language, and not have hearing impairment or generalised developmental delay. The diagnostic process of tongue-tie was based on functional and structural measures and completed by a lactation consultant and paediatric dentist before six months of age. All infants in the TTT and UTT groups were diagnosed with a functional restriction by an International Board Certified Lactation Consultant (IBCLC) combined with anatomical assessment by a paediatric dentist. Where a diagnosis of tongue-tie was made by the IBCLC, criteria included items listed in the Hazelbaker in addition to other symptoms, including milk/cell stasis on the dorsum of the tongue, aerophagia, milk transfer rates and others relevant to the infant. Inclusion criteria for participants in the UTT group were diagnosis of tongue-tie but no surgical treatment provided, while TTT children were diagnosed with tongue-tie and had frenulotomy by Erbium:YAG laser. NTT children had not been previously diagnosed with tongue-tie by any health professional.

A one-way between groups analysis of variance (ANOVA) and chi-square test of contingencies were used to assess group differences in age (months) and gender respectively. Assumptions were met for both tests and no significant difference was found for age, $F(2, 56) = 0.60$,

Table 1

Participant characteristics for the treated tongue-tie (TTT), untreated tongue-tie (UTT) and no tongue-tie (NTT) groups.

	n			Age (months)		
	Male	Female	Total	M	SD	Range
TTT	11	10	21	42.05	6.66	31–59
UTT	6	11	17	40.12	6.64	30–53
NTT	11	10	21	39.57	9.152	25–58
Total	28	31	59	40.61	7.59	25–59

Note. n = number of participants; M = mean, SD = standard deviation.

Table 2
Descriptive statistics on phonology, intelligibility, oral motor and tongue mobility for TTT, UTT and NTT groups.

		TTT			UTT			NTT		
		M	SD	R	M	SD	R	M	SD	R
Phonology	SS	9.29	3.39	3–16	8.29	2.09	3–12	8.52	2.99	3–13
	PCC	77.95	13.71	47–99	68.94	18.45	41–93	72.62	13.10	39–96
Intelligibility	Parent	4.05	.74	3–5	3.76	.75	2–5	3.86	.66	2–5
	Clinician	3.95	.87	2–5	3.53	.94	2–5	3.86	.91	2–5
VMPAC RS		31.67	3.47	24–36	31.71	3.69	24–36	31.60	4.06	23–36
Lingual frenulum RS		9.57	.75	8–10	8.88	1.27	7–10	9.71	.46	9–10

Note. M = mean; SD = standard deviation; R = range; SS = standard score; PCC = percentage consonants correct; VMPAC = Verbal Motor Production Assessment for Children; RS = raw score. Intelligibility scores are from a scale 1 to 5 (1 = completely unintelligible to 5 = completely intelligible).

$p = .55$, $\eta^2 = 0.021$ (small effect) or gender, $\chi^2(2, N = 59) = 1.42$, $p = .49$, $\Phi = 0.22$.

Sensitivity analysis conducted using G*Power software concluded that with 59 participants, there was 80% likelihood of achieving a minimum effect size of $f = 0.42$ (large effect) [35].

3. Materials

The following objective measures were included in the study.

3.1. Measures

3.1.1. Speech sound assessment

Depending on the age of the participant, either the Toddler Phonology Test (TPT) [36] or the phonology subtest of the Diagnostic Evaluation of Articulation and Phonology (DEAP) [37] was administered to assess each participant's speech sound production. Both tools are standardised assessments for speech sound production, contain normative data for Australian children, and use similar stimuli.

3.1.2. Intelligibility

Parent and clinician intelligibility ratings were used to measure the child's overall ability to be understood on a 5-point Likert scale [28,38].

3.1.3. Oral motor skills

Tongue mobility was assessed using the lingual control subtest of the Verbal Motor Production Assessment for Children [39].

3.1.4. Lingual frenulum function

To assess each child's current tongue-tie status, an assessment of lingual frenulum function for children over one year of age developed by Ito et al. was used [27]. A throat scope and permanent marker were used to measure item one objectively to the closest millimetre, by putting the throat scope underneath the child's tongue against the frenulum and marking the position of the tip of the tongue. Ito and colleagues [28] reported that items one to four were based on the Hazelbaker Assessment Tool of Lingual Frenulum Function (HATLFF) [40], which is considered a gold standard of lingual assessment with strong psychometric properties. The HATLFF, however, could not be used as it is designed for the assessment of infants under 12 months.

3.2. Procedure

Ethical approval was obtained from the Curtin University Human Research Ethics Committee. Written informed consent was obtained prior to the commencement of each session. Sessions of 30–45 min were completed at the participant's home in a quiet, distraction-free area, or at a private speech pathology clinic if requested by the family. Parents or guardians were permitted to observe and completed a parent questionnaire and intelligibility ratings during the session. The sequence of each session included: rapport building (clinician intelligibility rating

attained), speech production assessment, oral motor assessment, and lingual frenulum function assessment. A rest or reward activity was offered between tasks when necessary. All sessions were video-recorded with an iPhone and audio-recorded with an Olympus VN741PC recorder. Assessment data were transcribed after each session. Parents were provided with a summary of their child's assessment results if requested.

Two student speech pathologists (SSP), the chief investigator and an assistant, completed all assessments. To ensure inter-rater reliability between scorers, 20% of recordings (audio and video) were randomly selected and scored in their entirety by both SSPs. A high rate of consistency (> 95%) was achieved between the two raters, with any differences being discussed and agreement reached.

3.3. Data analysis

Statistical analyses were conducted using Statistical Package for the Social Sciences (SPSS) Version 23.0 software [41]. Outliers were not excluded as they captured the spread of participants.

4. Results

Descriptive statistics for speech sound (phonology and intelligibility), oral motor skills and lingual frenulum function (tongue mobility) are outlined in Table 2.

4.1. Speech sound assessment

Speech sound production skills were investigated through analysis of participants' phonology, measured by standard score and percentage of consonants correct (PCC) on the DEAP and TPT [36,37]. The assumption of normality was not met for all three groups for phonology standard score. The Kruskal-Wallis one-way ANOVA found no significant main effect for phonology standard score, $H(\text{corrected for ties}) = 2.63$, $df = 2$, $N = 59$, $p = .27$, $\eta^2 = 0.045$ (small-medium effect) between the three groups.

While groups were not significantly different by age (months), variations of age were present, therefore an ANCOVA with a covariate of age (months) for total PCC was completed. Normality assumption was violated for the UTT group ($p = .035$), but due to the robust nature of the ANCOVA and visual inspection indicating participants were normally distributed, this test was conducted. Other assumptions of homogeneity including regression, linearity and homogeneity of variance were not violated. After accounting for the effects of age on PCC, the ANCOVA found no significant effect of PCC between groups, $F(2, 55) = 1.36$, $p = .267$, partial $\eta^2 = 0.047$ (small-medium effect). The results indicated no significant difference between any groups on speech production measures.

Alveolar and palato-alveolar sounds were of particular interest given the impact of reduced tongue movement on the accuracy of production of these sounds, so these were investigated separately. The

assumption of normality was not met, therefore a Kruskal-Wallis one-way ANOVA was used. No significant difference was found between groups for the initial or final word position of alveolar (/t/,/d/,/s/,/z/,/n/,/l/) or palato-alveolar (/ʃ/,/ʒ/,/tʃ/,/dʒ/,/r/) sounds.

4.2. Intelligibility

Parent and clinician intelligibility ratings were compared between the TTT, UTT and NTT groups. As intelligibility ratings contain ordinal data, a non-parametric Kruskal-Wallis one-way ANOVA was used. No significant difference was found between groups for parent intelligibility rating, $H(\text{corrected for ties}) = 1.31$, $df = 2$, $N = 59$, $p = .52$, $\eta^2 = 0.023$ (small-medium effect) or clinician intelligibility rating, $H(\text{corrected for ties}) = 1.85$, $df = 2$, $N = 59$, $p = .40$, $\eta^2 = 0.032$ (small-medium effect).

4.3. Tongue mobility

Since the assumption of normality was not met for VMPAC scores between TTT, UTT and NTT groups, a Kruskal-Wallis one-way ANOVA was conducted. No significant difference was found between all groups for VMPAC raw score, $H(\text{corrected for ties}) = 0.028$, $df = 2$, $N = 58$, $p = .99$, $\eta^2 = 0.00049$ (no to small effect). Data for one participant in the NTT group was not available for analysis due to non-compliance on this task.

4.4. Tongue-tie status

Participants' current tongue-tie status' were classified on the basis of the lingual frenulum function scores [27]. The assumption of normality was not met for lingual frenulum function raw score; therefore, a Kruskal-Wallis one-way ANOVA was conducted. No significant difference was found between groups, $H(\text{corrected for ties}) = 5.24$, $df = 2$, $N = 59$, $p = .073$, $\eta^2 = 0.090$ (medium-large effect). Although no significant difference was found between lingual frenulum raw scores, a significant difference was detected for tongue-tie status, $H(\text{corrected for ties}) = 10.42$, $df = 2$, $N = 59$, $p = .005$, $\eta^2 = 0.18$ (large effect) between TTT (*Mean Rank* = 32, $n = 21$), UTT (*Mean Rank* = 25.06, $n = 17$) and NTT (*Mean Rank* = 32, $n = 21$) groups. This was analysed with a Kruskal-Wallis one-way ANOVA, as normality assumption was not met. Pairwise comparisons via Mann-Whitney U Test revealed a significant difference in tongue-tie status, between UTT (*Mean Rank* = 17.03, $n = 17$) and NTT groups (*Mean Rank* = 21.5, $n = 21$), $U = 136.5$, $z = -2.32$ (corrected for ties), $p = .020$, two-tailed, $r = 0.38$ (medium-large effect) and a significant difference between UTT (*Mean Rank* = 17.03, $n = 17$) and TTT (*Mean Rank* = 21.5, $n = 21$) groups, $U = 136.5$, $z = -2.32$ (corrected for ties), $p = .020$, two-tailed, $r = 0.38$ (medium-large effect). However, when Bonferroni correction was applied and the alpha level was adjusted to $p = .017$, pairwise comparisons indicated no significant difference between any of the pairs of groups (TTT and UTT, UTT and NTT, TTT and NTT).

4.5. History of speech pathology services

A Kruskal-Wallis one-way ANOVA revealed a significant difference in previous speech pathology services, $H(\text{corrected for ties}) = 6.54$, $df = 2$, $N = 59$, $p = .038$, $\eta^2 = 0.112$ (medium-large effect), between TTT (*Mean Rank* = 28.88, $n = 21$), UTT (*Mean Rank* = 25.82, $n = 17$) and NTT (*Mean Rank* = 34.50, $n = 21$) groups. Pairwise comparisons via the Mann-Whitney U Test with adjusted alpha level of $p = .017$, indicated a significant difference between UTT (*Mean Rank* = 6.41, $n = 17$) and NTT (*Mean Rank* = 22, $n = 21$) groups, $U = 126$, $z = -2.63$ (corrected for ties), $p = .008$, two-tailed, $r = 0.43$ (medium-large effect). The reasons for children attending speech pathology services can be seen in Table 3. No children in the NTT had previously received speech pathology services.

Table 3

Number of children in treated tongue-tie (TTT) and untreated tongue-tie (UTT) groups previously attending speech pathology for each goal of therapy.

Therapy Goals	TTT	UTT
Speech Sound Production	4 ^{*1}	2 ^{*2}
Oral Motor	0	1
Language	0	2
Unknown	0	1
Total n	4	5
% of group	19.05%	29.41%

Note. N = number of participants. One child in UTT group had received therapy for both speech and language.

^{*1} /l/,/s/,/r/,/ʃ/,/tw/,/θ/,/z/,/f/.

^{*2} /s/.

5. Discussion

This study investigated differences between speech production and tongue mobility across young children and found no significant difference between TTT, UTT and NTT groups on measures of speech production, parent and clinician intelligibility ratings and tongue mobility. Significantly more children in the UTT group had previously seen speech pathologists, in comparison to the NTT group. It was interesting to note that the majority of children within the UTT group no longer presented with a tongue-tie, as indicated by their lingual frenulum function score [27].

5.1. Tongue-tie and speech production

Although it is widely believed that tongue-tie can affect speech production [24,26,27 and 32], research in this area is inconclusive due to small sample sizes and variability in methodology and assessment tools used in existing studies [33]. In the current study, no significant difference in speech outcomes was found between children with UTT and NTT. These findings align with existing evidence of no causative effect of tongue-tie on speech production skills. Significant differences in speech production after surgery were reported in two studies [24,28], however the differences were based solely on parental reports and did not include objective measures of speech production. The current study compared children with TTT to UTT and a control group of NTT on a range of measures, including standardised speech assessments, parent and clinician intelligibility ratings, and found no significant difference in speech outcomes between the three groups. These results are similar to findings of a previous study by Dollberg and et al., who utilised a similar research design but with fewer participants, and found no significant difference in speech outcomes between TTT, UTT and NTT groups [32]. The current study did not support significant findings for speech production on the basis of parental report [24,28], with intelligibility not differing significantly between groups. However, previous studies that found this difference compared children's intelligibility pre- and post-surgery [24,28].

5.2. Tongue-tie and speech motor programs

In relation to the model of speech sensorimotor control [34], it was predicted that participants with UTT would need to create an alternative motor program to account for the impairment as a result of tongue-tie. It was believed that this altered motor program would create a difference in the sound of speech produced.

It was also hypothesized that TTT children would independently develop new motor programs, following the treatment of their tongue-tie, and would no longer present with alternative motor productions and difference in speech compared to NTT participants. Through auditory feedback of speech sounds produced by others and individual self-monitoring of speech, it was postulated that these children would

independently update motor programs. The main results from this study support this prediction, as the speech sound production scores of participants in the TTT group were not significantly different to NTT participants. However, further investigation into results revealed this hypothesis was not necessarily supported. When considering speech pathology history, 19% of children within the TTT group had seen a speech pathologist previously; all of whom had attended due to problems with alveolar and palato-alveolar sounds (/l/,/s/,/r/,/j/,/z/). This indicates that a high proportion of TTT children required assistance to develop new motor programs for sounds which required tongue movement, following their tongue-tie release. Therefore, although TTT children did not present with speech acoustically different to NTT children and appeared to have new motor programs, many needed support to create these programs.

It was anticipated that participants in the UTT group would not possess typical oral motor skills given the presence of tethered oral tissue, and it was predicted that these children would need to create alternative motor programs and thus present with altered speech production. However, there was no significant difference between groups on speech production outcomes, so this prediction was not supported. Although significantly more UTT children had seen speech pathology previously when compared to NTT children, only 12% of UTT children attended therapy for speech production, with 6% of the group specifying this was for an alveolar sound (e.g./s/). Most UTT children attended for other reasons, including language development and developing oral motor skills. This highlights that many UTT children independently learned to create sounds that were acoustically matched to the sounds of individuals without tongue-tie. Although speech sounds were not acoustically different, it would be interesting to conduct further analysis of speech sound productions of UTT children using a specialized speech analysis program such as Praat [42], or use of electropalatography, to measure contact with the palate and alveolar ridge during speech production. Perceptually, it appeared that participants in the TTT group needed support to create typical motor programs and the majority of UTT participants independently created motor programs which accounted for their tongue-tie. However, it is possible that participants in the TTT group had more restricted lingual frenulum structure before surgery than the UTT group, and that parental report of speech therapy goals may have been inaccurate.

5.3. Limitations and future directions

The Ito and colleagues lingual assessment tool [27] for children over one year of age was deemed the most appropriate assessment to measure current tongue-tie status, as it is the only assessment tool found for children over the age of one. However, throughout the study, the accuracy of the lingual frenulum tool was questioned, which may have impacted internal validity [27]. In particular, item five 'speech for age' contained no reliability data and Ito et al. [27] did not explain how this item was assessed, so it was unclear whether this score was measured based on speech sound errors, phonetic inventory, or intelligibility. The inclusion of this item within the assessment suggests there is an assumption that lingual frenulum function and tongue-tie status impact speech production. It was also interesting that the majority of children within the UTT group were not classified as having a tongue-tie, as indicated by this measure. This could be due to inadequate sensitivity of the lingual frenulum tool to diagnose tongue-tie within older children from two to five years of age. In the future, it would be beneficial to validate an accurate and reliable diagnostic tool of tongue-tie for children older than one year old.

All participants from the current study were recruited from a single clinic and received treatment with Erbium laser surgery. A direction for the future could be to replicate this study across multiple sites and with alternative surgical procedures, to determine if the results are generalised to a wider population. Secondly, the classification of tongue-tie, superior and inferior attachment and other functional

characteristics were not controlled in the current study, and tongue-tie severity may have differed between UTT and TTT groups. It is possible that UTT children had less severe tongue-ties, therefore, did not proceed with surgery. Noting the UTT and TTT cohort were initially seen for nursing dysfunction, the UTT group also included infants with functional and structural lingual frenulum dysfunction that were able to achieve adequate functional change with non-surgical interventions. This is a very important aspect to consider controlling in future prospective studies. Finally, it is suggested that the study be replicated with a larger sample size. It would also be beneficial to conduct a prospective study, which follows TTT, UTT and NTT participants over time, to track lingual frenulum structure and function, speech production, intelligibility, and tongue mobility.

6. Conclusion

There has been an increased awareness and surgical management of tongue-tie over the past decade [11,12]. Yet inconclusive evidence exists for the impact of tongue-tie presence on speech production in children, and current conclusions have been based on studies with small sample sizes, that lack control groups, and use of objective measures. The current study of 59 participants found that objective measures of speech outcomes, intelligibility and tongue mobility did not significantly differ between TTT, UTT and NTT groups. Further investigation accounting for this study's limitations are recommended. Due to the multidisciplinary nature of tongue-tie treatment, it is important that these findings are communicated to a range of health professionals to inform clinical decision making and allow sound advice to be provided to families about the impact of tongue-tie and the potential effects of on speech production and development [14,43,44].

References

- [1] C. Fernando, *Tongue-tie: from Confusion to Clarity*, Tandem, 1998 Sydney, Australia.
- [2] Mills N, Pransky SM, Geddes DT Mirjalili S. What is a Tongue Tie? Defining the anatomy of the in-situ lingual frenulum. *Sep;32(6):749-761*. doi: 10.1002/ca.23343. *Clinical Anatomy* 2019.
- [3] Katz RV, Dearing BA, Ryan JM, Ryan LK, Zubi MK, Sokhal GK. Development of a tongue-tie case definition in newborns using a Delphi survey: the NYU-Tongue-Tie Case Definition. *Oral Surg Oral Med Oral Pathol Oral Radiol*. S2212-4403(19) 30017-3. doi: 10.1016/j.oooo.2019.01.012.2019.
- [4] A. Haham, R. Marom, L. Mangel, E. Botzer, S. Dollberg, Prevalence of breastfeeding difficulties in newborns with a lingual frenulum: a prospective cohort series, *Breastfeed. Med.* 9 (9) (2014) 438–441.
- [5] L.M. Segal, R. Stephenson, M. Dawes, P. Feldman, Prevalence, diagnosis, and treatment of ankyloglossia: methodologic review, *Canadian family physician Medecin de famille canadien* 53 (6) (2007) 1027–1033.
- [6] J. Ballard, C. Auer, J. Khoury, Ankyloglossia: assessment, incidence, and effect of frenuloplasty on the breastfeeding dyad, *Pediatrics* 110 (5) (2002) 1–6.
- [7] L. Ricke, N. Baker, D. Madlon-Kay, T. DeFor, Newborn tongue-tie: prevalence and effect on breast-feeding, *J. Am. Board Fam. Pract.* 18 (1) (2005) 1–7.
- [8] A. Messner, M. Lalakea, J. Macmahon, E. Bair, Ankyloglossia: incidence and associated feeding difficulties, *Arch. Otolaryngol. Head Neck Surg.* 126 (1) (2000) 36–39.
- [9] M. Hogan, C. Westcott, M. Griffiths, Randomized, controlled trial of division of tongue-tie in infants with feeding problems, *J. Paediatr. Child Health* 41 (5–6) (2005) 246–250.
- [10] R.L.D.C. Martinelli, I.Q. Marchesan, G. Berretin-Felix, Posterior lingual frenulum in infants: occurrence and maneuver for visual inspection, *Revista CEFAC* 20 (4) (2018) 478–483.
- [11] J. Walsh, A. Links, E. Boss, D. Tunkel, Ankyloglossia and lingual frenotomy: national trends in patient diagnosis and management in the United States, 1997–2012, *Otolaryngology-Head Neck Surg. (Tokyo)* 156 (4) (2017) 735–740.
- [12] V. Kapoor, P.S. Douglas, P.S. Hill, L.J. Walsh, M. Tennant, Frenotomy for tongue-tie in Australian children, 2006–2016: an increasing problem, *Med. J. Aust.* 208 (2) (2018) 88–89.
- [13] A. Brookes, D.M. Bowley, Tongue-tie: the evidence for frenotomy, *Early Hum. Dev.* 90 (11) (2014) 765–768.
- [14] D.O. Francis, S. Krishnaswami, M. McPheeters, Treatment of ankyloglossia and breastfeeding outcomes: a systematic review, *Pediatrics* 135 (6) (2015) 1458–1466.
- [15] L.M. Segal, R. Stephenson, M. Dawes, P. Feldman, Prevalence, diagnosis, and treatment of ankyloglossia: methodologic review, *Canadian family physician Medecin de famille canadien* 53 (6) (2007) 1027–1033.
- [16] Carmela Baeza, Catherine Genna, James Murphy, Alison Hazelbaker,

- Martin Kaplan, Roberta Martinelli, Irene Marchesan, Pamela Douglas, Christina Smillie, Assessment and classification of tongue-tie, *Clinical Lactation* 8 (2017) 93–98, <https://doi.org/10.1891/2158-0782.8.3.93>.
- [17] P. Hong, D. Lago, J. Seargeant, L. Pellman, A. Magit, S. Pransky, Defining ankyloglossia: a case series of anterior and posterior tongue-ties, *Int. J. Pediatr. Otorhinolaryngol.* 74 (9) (2010) 1003–1006.
- [18] C. Genna, E. Coryllos, Breastfeeding and tongue-tie, *J. Hum. Lactation* 25 (1) (2009) 111–112.
- [19] Suter V, Bornstein M, Ankyloglossia: facts and myths in diagnosis and treatment., *J. Periodontol.* 8 (80) 1204–1219.
- [20] A. Srinivasan, C. Dobrich, H. Mitnick, P. Feldman, Ankyloglossia in breastfeeding infants: the effect of frenotomy on maternal nipple pain and latch, *Breastfeed. Med.* 1 (4) (2009) 216–224.
- [21] Victoria Bellinger, Donna Solari, Monica Hogan, Kathryn Rodda, Bruce Shadbolt, David Todd, Tongue-tie division in the newborn: follow-up at 9 and 38 months [online], *Breastfeed. Rev.* 26 (No. 1) (Mar 2018) 13–22. Availability <https://search.informit-com-au.dbgw.lis.curtin.edu.au/documentSummary;dn=522330841316583;res=IELHEA ISSN: 0729-2759>. [cited 22 Nov 19].
- [22] A.N. Webb, W. Hao, P. Hong, The effect of tongue-tie division on breastfeeding and speech articulation: a systematic review, *Int. J. Pediatr. Otorhinolaryngol.* 77 (5) (2013) 635–646.
- [23] J. Heller, J. Gabbay, C. O'Hara, M. Heller, J.P. Bradley, Improved ankyloglossia correction with four-flap Z-frenuloplasty, *Ann. Plast. Surg.* 54 (6) (2005) 623–628.
- [24] A.H. Messner, M.L. Lalakea, The effect of ankyloglossia on speech in children, *Otolaryngology-Head Neck Surg. (Tokyo)* 127 (6) (2002) 539–545.
- [25] M.L. Lalakea, A.H. Messner, Ankyloglossia: the adolescent and adult perspective, *Otolaryngology-Head Neck Surg. (Tokyo)* 128 (5) (2003) 746–752.
- [26] R. Baxter, L. Hughes, Speech and feeding improvements in children after posterior tongue-tie release: a case series, *International Journal of Clinical Pediatrics* 7 (3) (2018) 29–35.
- [27] Y. Ito, T. Shimizu, T. Nakamura, C. Takatama, Effectiveness of tongue-tie division for speech disorder in children, *Pediatr. Int.* 57 (2) (2015) 222–226.
- [28] A. Walls, M. Pierce, H. Wang, A. Steehler, M. Steehler, E.H. Harley Jr., Parental perception of speech and tongue mobility in three-year olds after neonatal frenotomy, *Int. J. Pediatr. Otorhinolaryngol.* 78 (1) (2014) 128–131.
- [29] P. Flipsen Jr., Speaker-listener familiarity: parents as judges of delayed speech intelligibility, *J. Commun. Disord.* 28 (1) (1995) 3–19.
- [30] Z.A. Camargo, I.Q. Marchesan, L.R. Oliveira, M.A.F. Svicerio, L.C.K. Pereira, S. Madureira, Lingual frenectomy and alveolar tap production: an acoustic and perceptual study, *Logoped. Phoniatr. Vocol.* 38 (4) (2013) 157–166.
- [31] S. Daggumati, J.E. Cohn, M.J. Brennan, M. Evarts, B.J. McKinnon, A.R. Terk, Speech and language outcomes in patients with ankyloglossia undergoing frenulectomy: a retrospective pilot study, *American Academy of Otolaryngology-Head and Neck Surgery* 3 (1) (2019).
- [32] S. Dollberg, Y. Manor, E. Makai, E. Botzer, Evaluation of speech intelligibility in children with tongue-tie, *Acta Paediatrica* 100 (9) (2011) 125–127.
- [33] S. Chinnadurai, D.O. Francis, R.A. Epstein, A. Morad, S. Kohanim, M. McPheeters, Treatment of ankyloglossia for reasons other than breastfeeding: a systematic review, *Pediatrics* 135 (6) (2015) 1467–1474.
- [34] A. Van der Merwe, A theoretical framework for the characterization of pathological speech sensorimotor control, in: M.R. McNeil (Ed.), *Clinical Management of Sensorimotor Speech Disorders*, second ed., Thieme Medical Publishers, New York, United States, 1997, pp. 3–18.
- [35] F. Faul, E. Erdfelder, A.G. Lang, A. Buchner, G*Power 3: a flexible statistical power analysis program for the social, behavioural and biomedical sciences, *Behaviour Research Methods* 39 (2007) 175–191 2007.
- [36] B. McIntosh, B. Dodd, *Toddler Phonology Test*, Pearson Education, Queensland, Australia, 2011.
- [37] B. Dodd, Z. Hua, S. Crosbie, A. Holm, *Diagnostic Evaluation of Articulation and Phonology*, Pearson Education, Queensland, Australia, 2002.
- [38] C. Allen, T.P. Nikolopoulos, D. Dyar, G.M. O'donoghue, Reliability of a rating scale for measuring speech intelligibility after pediatric cochlear implantation, *Otology & Neurology* 22 (5) (2001) 631–633.
- [39] D. Hayden, P. Square, *Verbal Motor Production Assessment for Children: Examiner's Manual*, The Psychological Corporation, San Antonio, TX, 1999.
- [40] A.K. Hazelbaker, *The Assessment Tool for Lingual Frenulum Function (ATLFF): Use in a Lactation Consultant Private Practice*, Doctoral dissertation, Pacific Oaks College, 1993.
- [41] IBM Corp, *IBM SPSS Statistics for Windows*, IBM, Armonk, NY, 2012 (Version 21.0) [Computer Software].
- [42] P. Boersma, D. Weenink, Praat: Doing Phonetics by Computer, *Phonetic Sciences*, Amsterdam, The Netherlands, 2016 (Version 6.0.20) [Computer software].
- [43] A.H. Messner, M.L. Lalakea, Ankyloglossia: controversies in management, *Int. J. Pediatr. Otorhinolaryngol.* 54 (2–3) (2000) 123–131.
- [44] A. Hathaway, R. McCauley, *Assessment and Management of Tongue-Tie in Children: A Survey of Related Professionals*, (2007) Retrieved from http://www.asha.org/Events/convention/handouts/2007/0530_Hathaway_Allyson_2.