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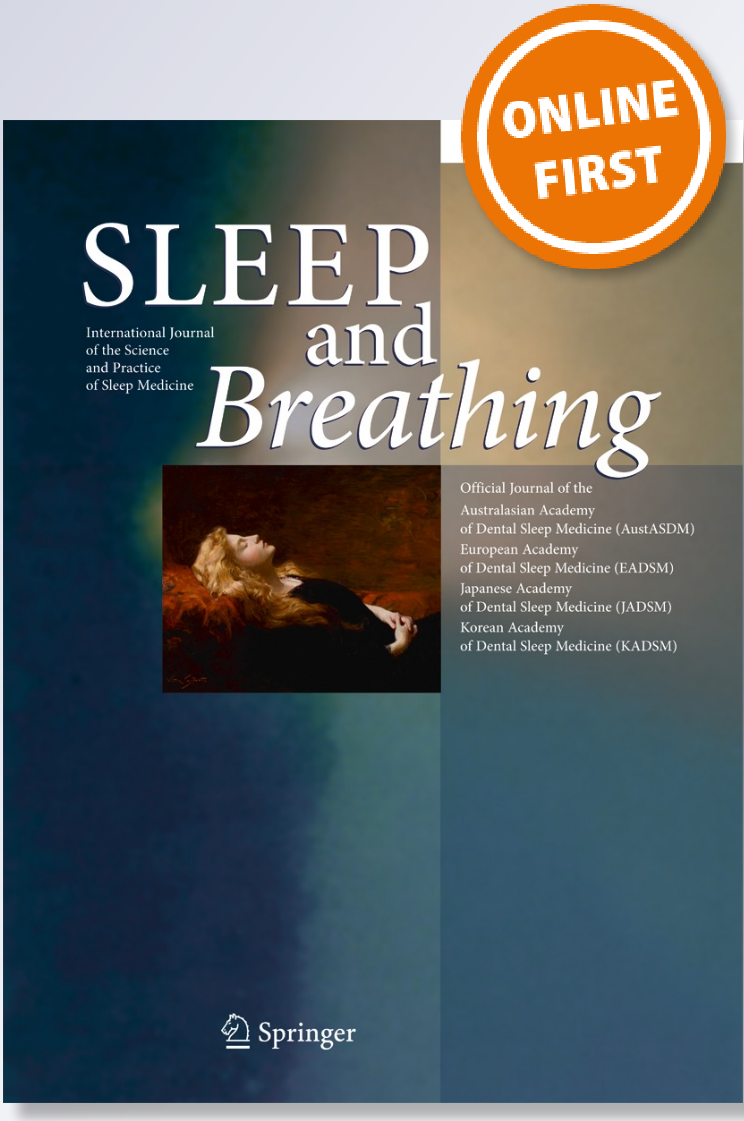

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Toward a functional definition of ankyloglossia: validating current grading scales for lingual frenulum length and tongue mobility in 1052 subjects

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Abstract
Purpose Alterations of the lingual frenulum may contribute to oromyofacial dysfunction, speech and swallowing impediments, underdevelopment of the maxillofacial skeleton, and even predispose to sleep breathing disorder. This study aims to assess the utility of existing instruments for evaluation of restricted tongue mobility, describe normal and abnormal ranges of tongue mobility, and provide evidence in support of a reliable and efficient measure of tongue mobility.
Methods A prospective cohort study of 1052 consecutive patients was evaluated during a 3-month period. Age, gender, ethnicity, height, weight, BMI, maximal interincisal mouth opening (MIO), mouth opening with tongue tip to maxillary incisive papillae at roof of mouth (MOTTIP), Kotlow's free-tongue measurement, and presence of severe tongue-tie were recorded. Secondary outcome measures include tongue range of motion deficit (TRMD, difference between MIO and MOTTIP) and tongue range of motion ratio (TRMR, ratio of MOTTIP to MIO).
Results Results indicate that MIO is dependent on age and height; MOTTIP and TRMD are dependent on MIO; Kotlow's free-tongue measurement is an independent measure of free-tongue length and tongue mobility. TRMR is the only independent measurement of tongue mobility that is directly associated with restrictions in tongue function.
Conclusions We propose the use of tongue range of motion ratio as an initial screening tool to assess for restrictions in tongue mobility. "Functional" ankyloglossia can thus be defined and treatment effects followed objectively by using the proposed grading scale: grade 1: tongue range of motion ratio is >80%, grade 2 50–80%, grade 3 < 50%, grade 4 < 25%.

Keywords Ankyloglossia · Frenulum · Tongue tie · Oromyofacial dysfunction · Classification of ankyloglossia · Tongue tie grading scale

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Introduction

The tongue is a dynamic organ that impacts breathing, speech, breastfeeding, and swallowing and thereby plays a critical role in facial development. In utero, the forward growth of the tongue is guided by the lingual frenulum, a thin strip of tissue that attaches the floor of the mouth to the ventral surface of the tongue [1]. During fetal development, the lingual frenulum functions to create a balance between the tongue, lip muscles, and growing facial bones. After birth, as the tongue muscles lengthen, the lingual frenulum retracts and becomes thin [2]. In some cases, the lingual frenulum fails to recede, tethering the tongue to the floor of the mouth. This results in ankyloglossia, commonly described as "tongue tie," a congenital oral anomaly that is characterized by an abnormally short or altered attachment of the lingual frenulum restricting tongue mobility to varying degrees [3].

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Ankyloglossia that functionally limits tongue mobility has tremendous clinical significance. It causes breastfeeding difficulties during infancy, including early weaning, diminished sucking and swallowing functions, and poor weight gain [4]. Ankyloglossia also affects upper and lower jaw development, periodontal tissue maturation, and dental occlusion. In a typically developing child, a balance exists between the lingual and the buccal musculature: the tongue pushes against the dentition and jaw in an expansile fashion, while the buccal musculature counters this force vector. With ankyloglossia, the tethering of the tongue to the floor of the mouth restricts adequate lingual force to expand the dentition and jaws. This leads to altered position and morphology of the jawbone [5]. Specifically, it has been shown that ankyloglossia contributes to maxillary hypoplasia (underdevelopment of the upper jaw bones), which then can predispose toward speech difficulty, nasal obstruction [6], mouth breathing [7], and obstructive sleep apnea [8].

It is important to identify patients with ankyloglossia at an early age, especially during facial development. When there are obvious nursing or speech difficulties in infants or young children, physicians more readily identify a severe lingual frenulum anomaly and direct patients to treatment [9]. However, differentiating the anatomical variations of the altered frenulum and the potential impact of mild to moderate restricted tongue mobility may be more challenging. Various methods to assess the degree of ankyloglossia and limitations in tongue mobility have been described [2, 4, 10–13]. Martinelli et al. have developed a comprehensive validated protocol for assessment of the lingual frenulum in infants that includes clinical history, anatomo-functional, and nutritive and non-nutritive suction evaluations [4, 14]. Similarly, Marchesan et al. have developed a similar protocol for assessment of the lingual frenulum in children and adults that includes clinical history, tongue mobility, anatomical shape, functional assessments, resting tongue position, and speech evaluations [10, 11]. These tools, however, require a time-intensive 30- to 60-min evaluation by a qualified and appropriately trained practitioner to administer. The objective of this study is to (1) validate the existing instruments for measurement of restricted tongue mobility in >1000 consecutive pediatric (6 years and older) and adult patients, (2) describe normal and abnormal ranges of restricted tongue mobility, and (3) propose a simple grading scale for the functional assessment of tongue mobility that is efficient for clinical use as a screening tool applicable in children and adults.

Methods

Study design

This is a prospective cohort study of 1052 consecutive patients evaluated in a private orthodontic office (AY) during the 3-month period from May 1, 2016 to August 1, 2016. Patients who participated in the study signed an informed consent form. Exclusion criteria included patients with a history of frenectomy and those with difficulty in mouth opening such as temporomandibular joint disorder. The institutional review board (IRB) of Stanford University approved the present study (protocol 35054, IRB no. 4947). Study data were collected and managed using Research Electronic Data Capture (REDCap) electronic data capture tools hosted at the Stanford Center for Clinical Informatics. REDCap [15] is a secure, web-based application designed to support data capture for research studies, providing (1) an intuitive interface for validated data entry, (2) audit trails for tracking data manipulation and export procedures, (3) automated export procedures for seamless data downloads to common statistical packages, and (4) procedures for importing data from external sources.

Data Collection method

The following data were prospectively and consecutively collected on all subjects who presented for orthodontic evaluation and provided written consent: age, gender, ethnicity (White, Hispanic, Asian, Indian, Black, Other), height (cm), weight (kg), BMI (kg/m^2), maximal interincisal mouth opening (MIO, mm), interincisal mouth opening with tongue tip to maxillary incisive papillae at roof of mouth (MOTTIP, mm), Kotlow's free-tongue measurement (length from base of tongue insertion of the lingual frenulum to the tip), and presence of severe clinically apparent ankyloglossia using Kotlow's structural guidelines. The quick-tongue tie assessment tool was used for measurements of MIO and MOTTIP, as well as Kotlow's free-tongue measurement (see Fig. 1).

All measurements were obtained with the patient in natural head position, which is a standardized and reproducible position of the head in an upright posture with the eyes focused on a point in the distance at eye level [16]. Natural head position implies that the visual axis is horizontal. For MIO measurement, patients were instructed to "open your mouth." The measurement was obtained on the first mouth opening to avoid jaw protrusion or excessive translation at the temporomandibular joint. Patients were not encouraged to open their mouth "as widely as possible." For MOTTIP measurement, patients were instructed to "touch the tongue to the back of the front two teeth and open your mouth." This measurement is obtained with the tongue at the incisive papillae and not at the incisive foramen which is used as "the spot" landmark during training with myofunctional therapy [17]. Kotlow's free-tongue measurement was obtained as previously reported [12] by measuring the length of the ventral surface of the tongue (while in full extension) from the insertion of the lingual frenulum to the tongue tip. The presence of severe clinically apparent tongue tie was assessed using Ruffoli's classification of levels of ankyloglossia and measurement

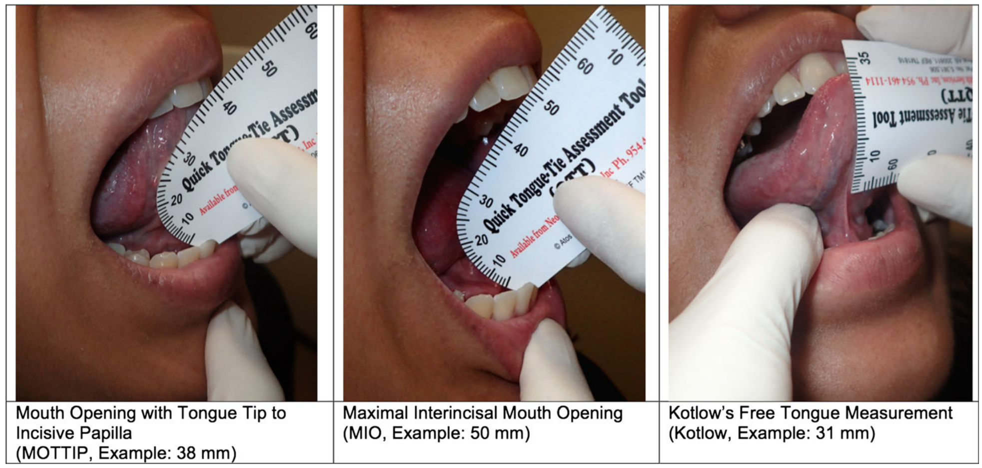

Fig. 1 Examples of tongue functioning and length measurements using the Quick Tongue Tie Assessment Tool (QTT): mouth opening with tongue tip to incisive papilla (MOTTIP), maximal interincisal mouth opening (MIO), and Kotlow's free-tongue measurement. Tongue range of motion deficit (TRMD) is defined as the difference between MIO and MOTTIP. Tongue range of motion ratio (TRMR) is defined as the ratio of MOTTIP to MIO

techniques A. The length of the frenulum was measured by recording the distance between the insertion of the lingual frenulum into oral floor and the tongue. If the length of frenulum was less than 7 mm, it was classified as "severe" in level of ankyloglossia [18].

Outcome measures

Primary outcome measures include MIO, MOTTIP, Kotlow's free-tongue measurement, and presence of severe clinically apparent tongue tie. Secondary outcome measures include tongue range of motion deficit (TRMD) calculated as the difference between the MIO and MOTTIP and tongue range of motion ratio (TRMR) calculated as the MOTTIP divided by MIO.

Statistical analysis

Statistical analyses were performed using JMP Pro 12 (SAS Institute Inc., Cary, NC). Continuous variables are summarized as mean (M) ± standard deviation (SD). Categorical variables are summarized as frequencies and percentages. Univariate analysis with Pearson's Chi square or independent *t* test (continuous variables) was performed to assess for nominal or continuous covariates of tongue measurements including age, gender, height, weight, BMI, and ethnicity. Bonferroni correction was applied to the interpretation of statistical significance due to the testing of multiple variables for each outcome, such that a two-tailed p value <0.0014 was required to achieve statistical significance.

Results

Our study included 1052 subjects with age ranging from 6 to 70 years. Demographic factors include age 20.1 ± 10.3 years (M ± SD); gender 61.7% female; height 162.5 ± 12.5 cm; weight 59.9 ± 17.8 kg; and BMI 22.4 ± 5.8 kg/m^2. Ethnicities include Hispanic 49.1%, Asian 25.8%, white 14.9%, non-Hispanic black 9.8%, and Indian 0.2%. This population includes 140 children (ages 6–11), 436 adolescents (age 12–17), 385 young adults (age 18–35), 84 adults (age 36–64), and 7 seniors (age > 65).

There were 40/1052 (3.8%) patients with severe clinically apparent tongue tie: 7 children, 14 adolescents, 10 young adults, and 9 adults. Measurements of tongue function of all patients are as follows (M ± SD): MIO 52.5 ± 5.4 mm, MOTTIP 33.6 ± 6.9 mm, Kotlow's free-tongue measurement 17.5 ± 5.5 mm, TRMD 18.0 ± 7.7 mm, and TRMR 64 ± 13%. The distribution of these measurements, including minimum, bottom 10% quantile, median, top 90% quantile, and maximum values for each age cohort are displayed in Fig. 2. Quantile box plots for each age cohort are displayed in Fig. 3. Visual assessment of the histograms for measurements of the overall population as shown in Fig. 2 demonstrates (1) MIO—multiple peaks, data symmetrical; (2) MOTTIP—multiple peaks, data skewed left; (3) Kotlow's measurement—single peak, data skewed right; (4) TRMD—multiple peaks, data skewed left; (5) TRMR—single peak, data skewed left, closely fits, and well modeled by the Johnson's S_U-distribution (a transformation of the normal distribution).

When compared to subjects without severe clinically apparent ankyloglossia, 40/1052 patients with severe clinical apparent ankyloglossia showed statistically significant differences in MOTTIP (22.2 ± 5.7 vs. 34.0 ± 6.5, $p < 0.0001$), Kotlow's measurement (12.0 ± 3.8 vs. 17.1 ± 3.8, $p < 0.0001$), TRMD (31.5 ± 7.2 vs. 18.4 ± 7.3, $p < 0.0001$), and TRMR (0.42 ± 0.10 vs. 0.65 ± 0.12, $p < 0.0001$).

Multivariate analysis with Standard Least Squares model shows *age* and *height* to be significant covariates of *MIO* (age: beta −0.08 ± 0.02, $p < 0.001$ and height: beta 0.14 ± 0.03, $p < 0.001$); the effect of these variables was strongest among children and adolescents under 18 years of age (age: beta 0.59 ± 0.08, $p < 0.001$ and height: beta 0.19 ± 0.02,

Cohort	All Patients	Children	Adolescents	Young Adults	Adults	Seniors
Ages (years)	6 - 70	6 - 11	12 - 17	18 - 35	36 - 64	> 65
N	1052	140	436	385	84	7
Maximum Interincisal Mouth Opening (mm)						
Mean	52.5	49.4	53.1	53.1	52	48.4
Std Dev	5.4	5.3	5.2	4.8	5.9	11.4
Min	32	38	38	32	39	33
10% Quantile	46	42	47	48	44.5	33
Median	53	50	53	53	52	53
90% Quantile	60	55	60	60	60	60
Max	70	70	70	65	65	60
Mouth Opening: Tongue Tip to Incisive Papilla (mm)						
Mean	33.6	31	33.1	35.1	33.5	33.7
Std Dev	6.9	6.8	7	6.4	7.2	6
Min	3	13	10	3	8	25
10% Quantile	25	22	23	27	24.5	25
Median	35	31	33	36	35	33
90% Quantile	42	40	42	43	43	42
Max	52	45	52	50	45	42
Kotlow's Measurement (mm)						
Mean	17.5	16.3	16.9	18.6	18	16
Std Dev	5.5	4.4	5.2	6.1	4.9	2.4
Min	2	7	6	2	5	13
10% Quantile	12	11	11	13	13	13
Median	16	16	16	17	17.5	16
90% Quantile	25	22	23.4	26	24.5	20
Max	40	35	35	40	34	20
Tongue range of motion deficit (mm)						
Mean	18.9	18.4	20.1	18.0	18.5	14.7
Std Dev	7.7	7.8	7.9	7.0	8.7	8.4
Min	4	4	5	5	5	5
10% Quantile	10	10	11	10	9.5	5
Median	18	17	19	17	17.5	17
90% Quantile	30	29	32	27	34	28
Max	53	42	45	53	40	28
Tongue range of motion ratio (%)						
Mean	64%	63%	63%	66%	65%	71%
Std Dev	13%	14%	13%	12%	15%	12%
Min	5%	24%	24%	5%	17%	53%
10% Quantile	46%	44%	43%	51%	42%	53%
Median	66%	65%	64%	68%	67%	70%
90% Quantile	80%	80%	79%	80%	81%	88%
Max	91%	90%	90%	91%	90%	88%

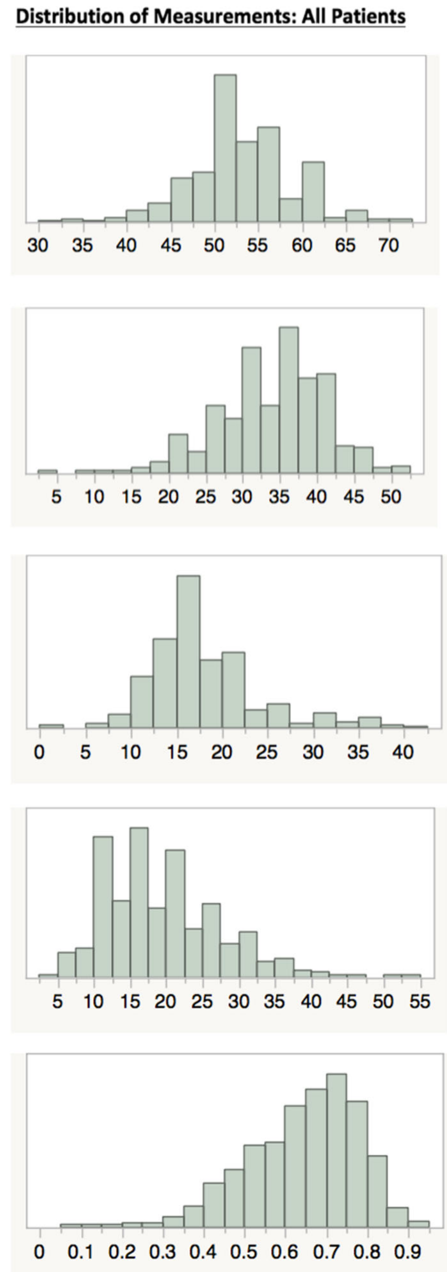

Fig. 2 Distribution of mouth opening, tongue measurements, tongue range of motion deficit, and tongue range of motion ratio by age cohort and overall population

$p < 0.001$). See Fig. 4a, b. *Age* was also a statistically significant covariate of *TRMD* (beta -0.09 ± 0.03, $p = 0.0012$). *Kotlow's free-tongue measurement*, *MOTTIP*, and *TRMR* were independent of age, gender, ethnicity, height, weight, and BMI. *Kotlow's free-tongue measurement* and *TRMR* were independent of *MIO*, whereas *MOTTIP* and *TRMD* had significant linear correlation with the *MIO* variable (R^2 0.05 and 0.25, respectively, $p < 0.0001$). *Kotlow's free-tongue measurement* and *TRMR* correlated with each other more strongly among patients with severe ankyloglossia (R^2 0.485) than those without severe ankyloglossia (R^2 0.314, $p < 0.001$). See Fig. 5.

Discussion

Tongue position and mobility play significant roles in facial skeletal development. Patients with aberrant development of

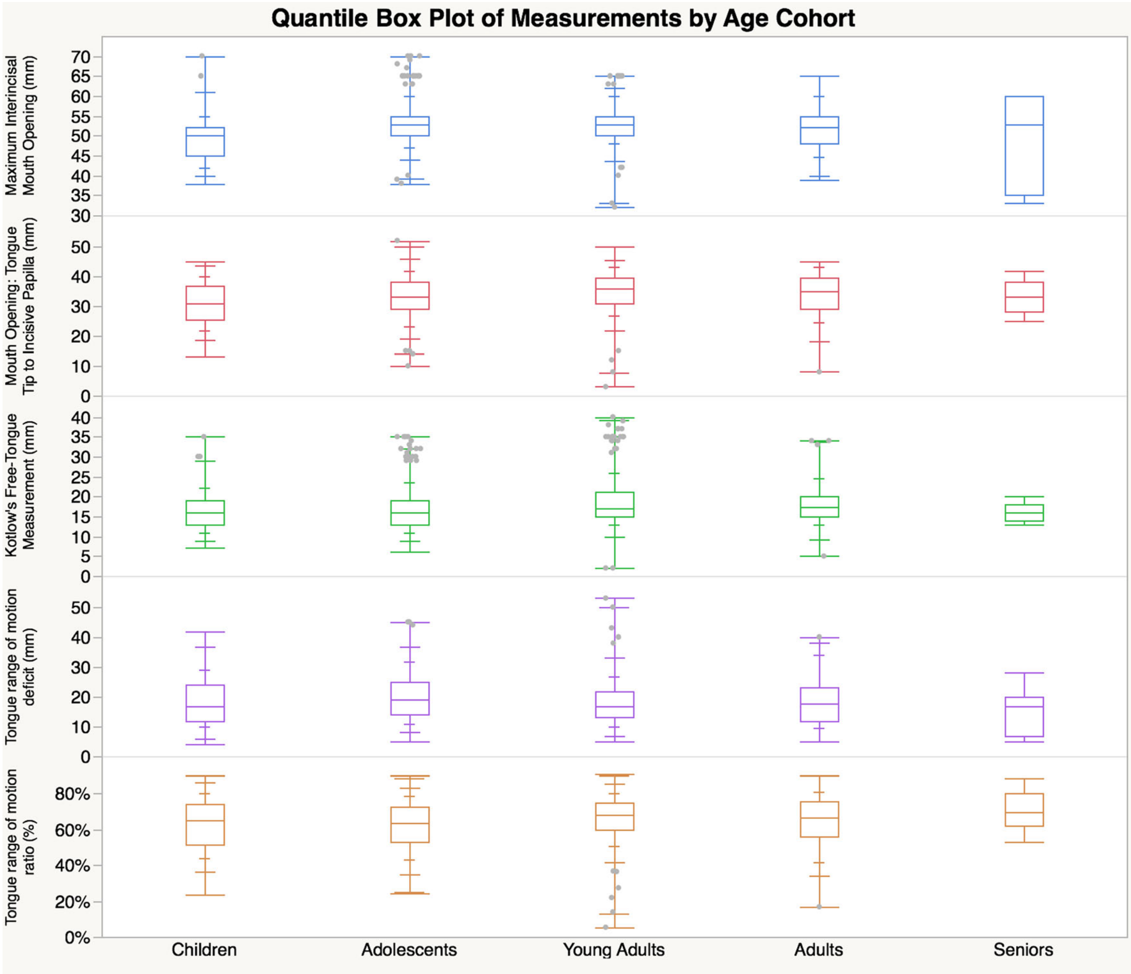

Fig. 3 Quantile box plot of measurements by age cohort: MIO, MOTTIP, Kotlow's free-tongue measurement, TRMD, and TRMR. Children 6–11 years; adolescents 12–17 years; young adults 18–35 years; adults 36–64 years; seniors >65 years

the upper or lower jaws are at increased risk of malocclusion [19], temporomandibular disorders [20], nasal obstruction [7], and obstructive sleep apnea [21]. Most recently, there have been concerted research efforts to explore the role of ankyloglossia and restricted tongue mobility as correctable risk factors of nasal obstruction and sleep-disordered breathing [17, 21, 22].

While the lingual frenulum and tongue mobility have come to the attention of the sleep medicine academic community, there is presently limited published data to guide the differentiation between normal and abnormal ranges of tongue mobility [10, 12, 18]. Here, we built upon the existing tools for assessment of tongue mobility by describing normal and abnormal parameters of tongue function among >1000 consecutive pediatric and adult subjects and provide evidence of a reliable measure of tongue mobility that is quick to use for all clinicians.

Kotlow et al. recognized the need for a classification system of ankyloglossia (tongue tie) over 15 years ago and proposed the free-tongue measurement to identify abnormal lingual frenulum attachments in the pediatric population [12]: 322 children with ages 18 months to 14 years were evaluated, where Boley gauge was used to measure the distance from the tip of the tongue to the insertion of the lingual frenulum. The study proposed that the normal length would be greater than 16 mm. Ankyloglossia was classified as Class I (mild, 12 to 16 mm), Class II (moderate, 8 to 11 mm), Class III (severe, 3 to 7 mm), and Class IV (complete ankyloglossia). A weakness with this method is that the tongue is especially flexible in young children and is difficult to control and stabilize during measurement [12]. To mitigate this weakness, structural guidelines were also used to assist in the determination of functional tongue limitations. Ruffoli et al. followed with a validation study in 200 children aged 6 to 12 years to describe normal and abnormal ranges of frenulum measurement (normal ≥ 20 mm, mild ankyloglossia 16–19 mm, moderate 8–15 mm, severe ≤ 7 mm) as well as MOTTIP (normal ≥ 23 mm, mild ankyloglossia 17 to 22 mm, moderate 4 to 16 mm, severe ≤3 mm) [18].

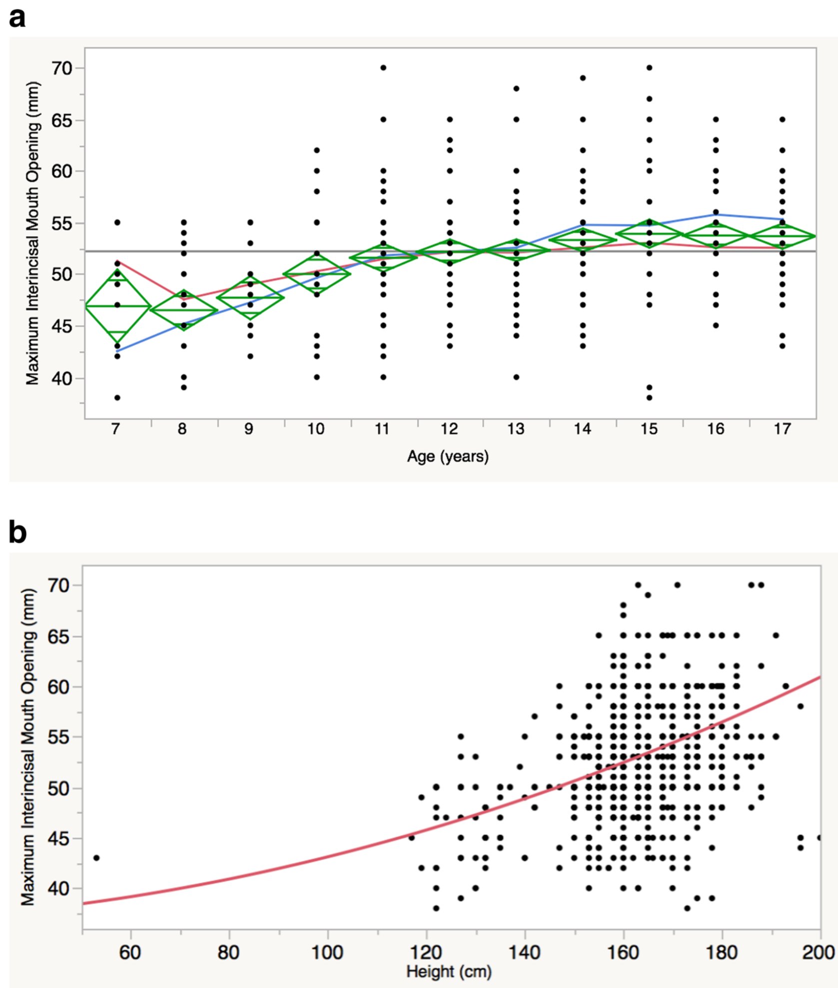

Fig. 4 Measurement of maximal interincisal mouth opening was found to be dependent on patient age and height. **a** Maximum interincisal mouth opening (mm) by age (years). The *red line* represents female gender, the *blue line* represents male gender, and the *green line* shows the mean diamond including the mean with 95% confidence interval of the mean for each age in years. **b** Maximum interincisal mouth opening (mm) by height (cm) for age <18 years

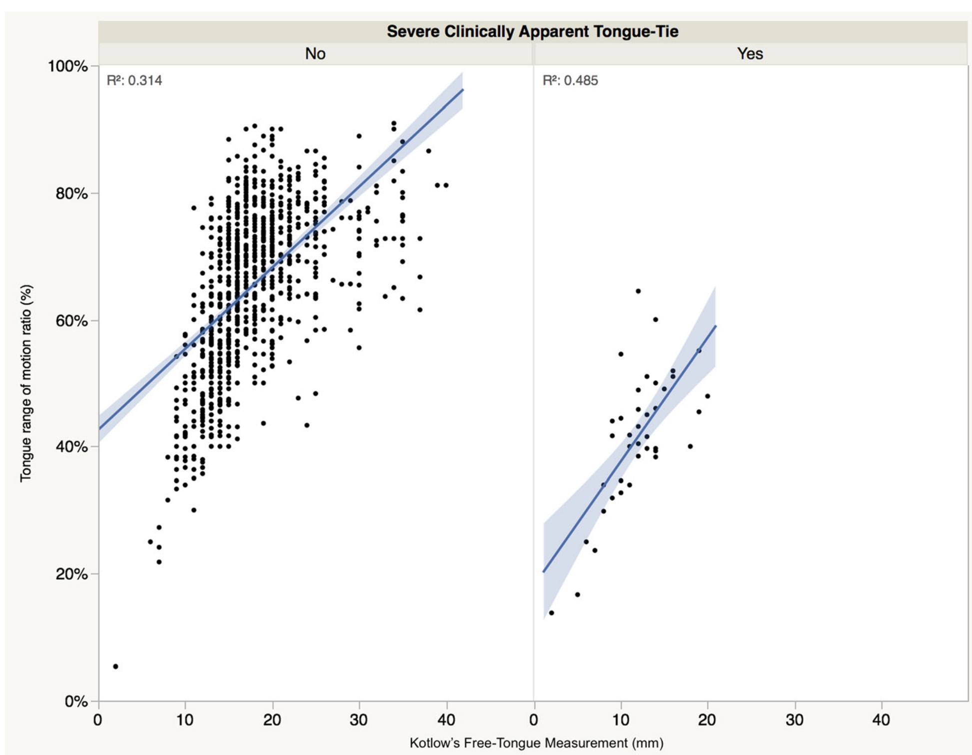

Fig. 5 Kotlow's free-tongue measurement vs. assessment of functioning with tongue range of motion ratio (TRMR). Kotlow's free-tongue measurement correlates only modestly with tongue range of motion (functioning). Kotlow's free-tongue measurement <20 mm was found to have 16.1% sensitivity and 77.6% specificity as a tool to predict below average tongue functioning in this series (TRMR grades 3–4). Among 295/1052 (28.0%) subjects with Kotlow's free-tongue measurements in the normal range (> 20 mm), there were still found to be 66 subjects with below average tongue functioning

Hazelbaker et al. developed a lingual frenulum measurement for use in breast-feeding infants. The Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) is based on clinical observations from lactation counselors [13]. HATLFF consists of five structural and seven functional criteria that have been validated by multiple groups with moderate reliability. [23–25] However, HATLFF only applies to infants.

Marchesan et al. developed a quantitative method to classify the lingual frenulum in adults for assessment of speech-language pathology [10]. Based on 98 adult subjects, they characterized the relationship between maximal interincisal mouth opening (MIO) to tongue tip movement toward the incisal papilla (TTIP). This is described either by (1) the difference between MIO and TTIP (TRMD) or (2) the ratio of TTIP to the MIO (TRMR). Among 16 subjects with short frenulum and 82 patients with normal frenulum, MIO was 46.6 ± 5.1 vs. 47.9 ± 6.9 mm (NS); MOTTIP was 28.1 ± 4.7 vs. 33.1 ± 5.0 (p = 0.0005); TRMD was 19.4 ± 7.7 vs. 13.4 ± 6.1 (p = 0.0056); and TRMR was 50 vs. 60%. A limitation of this study in adults was the sample size. Marchesan et al. then followed up with the same protocol to assess the lingual frenulum in more than 1400 adults [11] and infants [4] age 8 months to 62 years. [26] This allowed for the development of a comprehensive qualitative tool. [11].

To date, our study is the largest quantitative validation of Kotlow's free-tongue measurement and Marchesan's MOTTIP, TRMD, and TRMR measurements in 1052 subjects with age ranging from 6 to 70 years. Our results indicate that (1) MIO is dependent on age and height, especially for children and adolescents under 18 years of age (consistent with prior reports [27]). Hence, the covariates age and height need to be considered when determining quantile cutoffs for MOTT and TRMD as these measurements are dependent on MIO; (2) Kotlow's free-tongue measurement and Marchesan's TRMR are independent measures of free-tongue length and tongue mobility respectively and are not significantly influenced by age, gender, height, weight, ethnicity, or MIO; (3) Kotlow's free-tongue measurement has low sensitivity (16.1%) and only modest specificity (77.6%) for the diagnosis of functional ankyloglossia (tongue-tie); and (4) Marchesan's TRMR is the only independent measurement of tongue mobility that is directly associated with restrictions in tongue function.

Limitations to the study include (1) unblinded single-rater measurements based on one clinical encounter, (2) disproportionate racial ethnic groups (although this can be a strength as compared to previous studies which had more homogeneous ethnic populations), (3) cross-sectional nature of the study (where long-term longitudinal follow-up studies may be most ideal but not feasible), (4) no patients under the age of 6, and (5) analysis based on subjective "clinically significant" ankyloglossia.

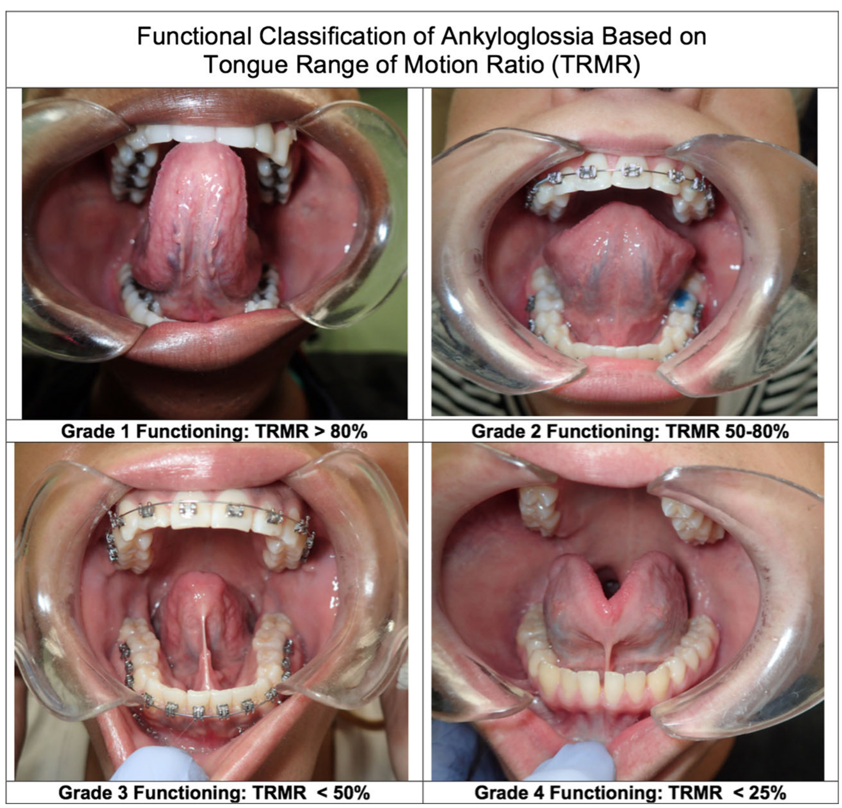

Fig. 6 A grading scale for the functional classification of ankyloglossia is proposed based on the TRMR (ratio of MOTTIP to MIO) building on the classification of Ferrés-Amat et al. [28]. Grade 1: tongue range of motion ratio is >80%, grade 2 50–80%, grade 3 <50%, grade 4 <25%. Higher grades reflect decreased tongue mobility and increased severity of tongue tie. The *photos* here demonstrate the deficit in the mobility of the tongue tip relative to MIO. With increasing ankyloglossia, the tongue tip is unable to touch the incisive papilla unless the mouth opening is closed to some extent. Considering grade 3, mouth opening is limited to 50% of maximal opening in order for the tongue tip to reach the incisive papilla. For grade 4, mouth opening is limited to 25% of MIO for the tongue tip to reach the incisive papilla

Conclusion

We propose the use of TRMR as an initial screening tool to assess for restrictions in tongue mobility, where a normal value for TRMR is between 51 and 77% (this represents the M ± 1 SD to include 68% of the population). Values below 46% can be considered significantly below average (bottom 10%), and values greater than 80% represent significantly above average functioning (top 10%). A functional TRMR grading scale based on our findings is proposed in Fig. 6: grade 1 = >80%, grade 2 = 50–80%, grade 3 = <50%, grade 4 = <25%. With the high reliability and precision of TRMR in assessing tongue mobility, our proposed grading scale enables a functional definition of ankyloglossia that can be used to assess treatment effects of myofunctional therapy and frenulum surgery. The grading scale allows clinicians to effectively communicate tongue mobility and associated nasal and oral function as they pertain to facial development. Further studies are needed to characterize the specific alterations to the lingual frenulum that render functional impairments.

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Author contributions All the authors met the four criteria for authorship established by the International Committee of Medical Journal Editors: Audrey Yoon, Soroush Zaghi, and Stanley Liu were responsible for the conception, design, analysis; drafting and revising the work; and reviewing the manuscript. Rachel Weitzman and Sandy Ha had substantial contributions to the acquisition of data for the work as well as in drafting and revising the work and reviewing the manuscript. Clarice S. Law and Christian Guilleminault had substantial contributions to data analysis, interpretation of data for the work, and revising the work critically for important intellectual content. Additionally, all authors provided final approval of the version to be published and agreed to be accountable for all aspects of the work including ensuring the accuracy and/or integrity of the work.

Compliance with ethical standards

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Conflict of interest The authors declare that they have no conflict of interest.

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Comments

Otolaryngologists and pediatricians pay close attention to the frenulum status of newborns to enable breastfeeding. Dentists can evaluate for functional tongue tie and contribute to proper growth and development of the maxilla. This article gives us an easy way to make a judgment, but it mostly will help the dentist pay attention.

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