THE Laryngoscope
FOUNDED IN 1896

Lingual frenuloplasty with myofunctional therapy: Exploring safety and efficacy in 348 cases.

Journal:	*The Laryngoscope*
Manuscript ID	Draft
Wiley - Manuscript type:	Original Reports
Date Submitted by the Author:	n/a
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Keywords - Combo:	Snoring < Sleep Medicine, Sleep < Pediatric airway < Pediatrics, Speech language pathology < Laryngology, Quality of Life < Laryngology, Surgical treatment of Obstructive sleep apnea < Sleep Medicine

Lingual frenuloplasty with myofunctional therapy: Exploring safety and efficacy in 348 cases.

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Acknowledgement: We would like to acknowledge the contributions of Chad Knutsen, Jennifer Rodriguez, William Belshe, Maryam Norouz, Katie Romero, Shahryar Barzegar, Debbie Hang, and Nora Ghodousi-Zaghi for their collaboration in development of this manuscript. No sponsorship or external funding was used to support this study.

Disclaimer: For author Macario Camacho, it is noted that the views herein are the private views of the author and do not reflect the official views of the Department of the Army or the Department of Defense.

Short running title: Lingual frenuloplasty with myofunctional therapy.

Number of tables and figures: 8
Numbers of pages for the entire manuscript: 19
Word Count: 4155

Abstract

Background: Ankyloglossia (tongue-tie) is a condition of altered tongue mobility due to the presence of restrictive tissue between the undersurface of the tongue and the floor of mouth. Potential implications of restricted tongue mobility (such as mouth breathing, snoring, dental clenching, myofascial tension) remain underappreciated due to limited peer-reviewed evidence. Here we explore the safety and efficacy of lingual frenuloplasty and myofunctional therapy for the treatment of these conditions in a large and diverse cohort of patients with restricted tongue mobility.

Methods: 420 consecutive patients (ages 29 months to 79 years) treated with myofunctional therapy and lingual frenuloplasty for indications of mouth breathing, snoring, dental clenching, and/or myofascial tension were surveyed. All procedures were performed by a single surgeon using a scissors and suture technique. Safety and efficacy was assessed >2 months post-operatively by means of patient-reported outcome measures.

Results: 348 surveys (83% response rate) were completed showing 91% satisfaction rate and 87% rate of improvement in quality-of-life through amelioration of mouth breathing (78.4%), snoring (72.9%), clenching (91.0%) and/or myofascial tension (77.5%). Minor complications occurred in < 5% of cases including complaints of prolonged pain or bleeding, temporary numbness of the tongue-tip, salivary gland issues, minor wound infection or inflammation, and need for revision to excise scar tissue. There were no major complications.

Conclusion: Lingual frenuloplasty with myofunctional therapy is safe and potentially effective for the treatment of mouth breathing, snoring, clenching, and cervico-facial tension in appropriately selected patient candidates. Further studies with objective measures are merited.

Level of Evidence: 2b

Keyword: lingual frenuloplasty, tongue-tie, lingual frenum, frenectomy, ankyloglossia, myofunctional therapy, orofacial myology, tongue and orofacial exercises.

Introduction

Ankyloglossia (also known as tongue-tie) is a condition of altered tongue mobility due to the presence of restrictive tissue in the midline between the undersurface of the tongue and the floor of mouth [1,2]. Restricted tongue mobility may be caused by a short mucosal lingual frenulum (commonly known as "anterior" tongue-tie) and/or by sub-mucosal myofascial fibers of the underlying genioglossus muscle that are fibrosed and impair optimal oral functions (also known as "posterior" tongue-tie) [3,4]. Ankyloglossia may also be attributed to scar tissue from a prior surgical procedure or other trauma.

The un-tethered mobility of the tongue is required for optimal speech, chewing, swallow, oral hygiene, and breathing functions [2,5], as well as for development of the maxillofacial complex and upper airway [6, 7]. Because the tongue plays such an important role in so many functions, restricted mobility of the tongue muscle may lead to dysfunctional compensations that may negatively affect nasal breathing and snoring due to low tongue posture or weigh on the other muscles of the face, mouth, neck, and shoulders. Moreover, the tongue is directly connected to the hyoid bone and has connections to the whole body (through the fascial diaphragms all the way down to the feet) through webs of connective tissue known as fascia [8,9]. A restrictive tongue may place tension on the deep front line of fascia (among other connective tissue networks) and contribute to neck tension, pain, and postural dysfunction [2,10]. As such, compensations for ankyloglossia may contribute to a wide variety of issues presenting as oral myofascial dysfunction.

Orofacial myofunctional therapy (also known as orofacial myology) has been used for many years to re-pattern and improve the function of the oral and facial muscles and to eliminate oral habits, such as prolonged thumb-sucking and nail biting, tongue thrusting, open-mouth at rest posture, incorrect mastication, and poor oral rest postures of the tongue and lips [11]. More recently, myofunctional therapy has been demonstrated as a potentially effective treatment option for snoring [12] and obstructive sleep apnea [13], and may soon be recognized as the most ideal initial treatment option for sleep-disordered breathing, [14] especially among pediatric populations [15].

However, restricted tongue mobility may interfere with the goals and limit the efficacy of myofunctional therapy. Patients with ankyloglossia may experience difficulty protruding, lateralizing, and most importantly elevating the tip or body of the tongue. Such functional impairments in the mobility of the tongue may prove a barrier in achieving tongue-to-palate contact necessary to create the "suction-cup" effect that holds the tongue in place and prevents it from falling into the pharynx. The purpose of the present study is to assess the safety and efficacy of lingual frenuloplasty in helping patients optimize the

efficacy of myofunctional therapy for the treatment of oromyofascial dysfunction in a large cohort of adult and pediatric patients.

Methods

Study Design

This is a retrospective cohort study of 420 consecutive patients who underwent lingual frenuloplasty as an adjunct to myofunctional therapy for the treatment of restricted tongue mobility and oromyofascial dysfunction associated with symptoms of mouth breathing, snoring, low tongue posture, tongue thrust, bruxism, swallowing issues, and/or cervico-facial tension. All procedures were performed by a single Otolaryngologist - Sleep Surgeon (SZ) at *The Breathe Institute*. Myofunctional therapy was performed by 45 different therapists of various levels of skill and expertise (under the direction and supervision of SZ and/or SVP) all of whom were trained and practice in the United States. The study involved a retrospective chart review and telephone survey of patients treated between March 12, 2016 to May 2, 2018. Verbal informed consent was obtained to participate in the survey. The study was performed as part of Stanford University IRB Number 6208, Protocol # 36385 approved on January 25, 2016.

Inclusion Criteria

Patients older than 2 years of age who were treated with myofunctional therapy and lingual frenuloplasty were invited to participate in the survey. Patients who were treated with lingual frenuloplasty in combination with other surgical procedures (such as adenoidectomy, tonsillectomy, or septoplasty) were excluded.

Treatment Protocol - Myofunctional Therapy

The goal of tongue-tie release in children, adolescents, and adults is to establish tongue tone, habituate correct posture, and enhance mobility; the tongue should maintain continuous contact with the roof of the mouth at rest and normalization of a mature lingual-palatal swallow must be achieved at the completion of treatment. As such, pre and post-operative oral myofunctional therapy is essential for optimal preparation and recovery after tongue-tie surgery. All patients treated with lingual frenuloplasty at *The Breathe Institute* were required to complete at least one month of pre-operative and two months of post-operative myofunctional therapy. The goals of pre-operative therapy are to create awareness of oral posture and tongue functions, strengthen and tone the muscles of the tongue and orofacial complex, and rehabilitate compensation patterns that may affect the post-operative recovery (e.g. floor of mouth elevation, muscular neck engagement, inability to perform isolated movements with the tongue without moving the jaw). Post-operative myofunctional therapy for lingual frenuloplasty provides individualized

care for the patient to optimize recovery and healing after surgery by providing guidance with passive and active wound stretching, as well as strength training and pattern retraining exercises for the tongue and orofacial muscles. Myofunctional therapy often continues for one year or longer to prevent relapse of dysfunctional oral motor habits, promote exclusive nasal breathing, and ensure long-term habituation of ideal resting oral posture. In addition to myofunctional therapy, many patients also received hands-on manual therapy in the form of myofascial therapy, craniosacral therapy, osteopathy, orthopedic physical therapy, massage, and/or chiropractic therapy depending on the clinical circumstance. Addressing compensatory muscular and joint tension through manual therapy before and after surgery helps to optimize rehabilitation and improves dysfunctional postural patterns and habits that have developed as a functionally compromised compensatory behaviors accommodating myofascial lingual frenulum restrictions.

Lingual Frenuloplasty with Scissors and Suture Technique-

Local anesthesia is achieved by applying topical viscous lidocaine followed by 0.5-1.7 cc of 1% lidocaine with 1:200000 epinephrine to the lingual frenulum via a 27-gauge needle. The patient is instructed to open the mouth and hold the tip of the tongue to the incisive papilla behind the maxillary central incisors to reveal tension to the lingual frenulum band. Tension is applied to the floor of the mouth with a groove director so as to protect the floor of mouth salivary glands. A hemostat is used to clamp the restrictive lingual frenulum 2-5 mm above the attachments of the submandibular gland duct. The mucosal frenulum is gently excised with the use of 120 mm Baby Metzenbaum or Iris scissors (curved or straight tip). The median lingual septum (fascia between the two head of the superior branch of the genioglossus muscle) is identified and dissected. The underlying myofascial fibers of the genioglossus muscle are dissected further with a combination of blunt and sharp dissection. Sterilized blunt cotton-tips and manual palpation with 2x2 sterile cotton gauze are used for blunt dissection. The patient undergoes a myofunctional assessment intra-operatively to determine for the presence of residual restrictive muscle or fascia bands that are restrictive of tongue mobility which are then excised sharply with scissors. The dissection is continued until adequate improvement to tongue mobility is achieved: i.e., tongue could be extended up towards the maxillary central incisors in maximal mouth opening position as well as held in lingual-palatal suction against the entire anterior and posterior aspects of the roof of the mouth without tension or strain. For cases performed under general anesthesia, a 2-0 silk suture is applied and used to mobilize the tongue for similar movements. Simple interrupted 3-0 or 4-0 chromic sutures are used to close the diamond-shaped mucosal defect and promote healing by primary intention healing. There was no use of electrocautery, silver nitrate, or thermal ablation with laser in this scissors and suture technique for lingual frenuloplasty. Hemostasis was achieved with suture ligation techniques and/or application of 2x2 gauze

until bleeding subsided. No antibiotics were prescribed or administered post-operatively. Patients were recommended oral rinses with salt water or non-alcoholic mouthwash three times daily for 1-2 weeks after the procedure; some patients also elected to use Vitamin E oil or colloidal silver spray. Pain control regimen included application of topical 2% viscous lidocaine, ibuprofen, Tylenol, and/or narcotics such as tramadol, hydrocodone, or oxycodone (as needed for more severe pain). Some patients elected to use homeopathic (such as arnica) or holistic remedies (turmeric, ginger, cannabidiol oil) for analgesia instead of the other more routine allopathic medications.

Survey

There were 348 surveys completed among 420 consecutive patients who were contacted to participate in the study (83% response rate). The patient surveys were conducted at least two months after the frenuloplasty procedure in a structured interview format by one of three research assistants who were not clinically involved in patient care. Patients were surveyed on the effects following treatment on their sleep, breathing, speech, and swallow patterns. The following items were assessed: age (years); gender (male vs. female); height (inches); weight (pounds); tongue tie severity (grades one through four using the functional classification of ankyloglossia based on the tongue range of motion ratio) [5]; indication for lingual frenulum release; local vs general anesthesia used for the procedure; date of procedure and length of time to follow-up. Incidence and severity of any complications including: pain, bleeding, numbness, and/or salivary gland issues was graded based on a one to ten-point visual analogue scale. Changes to the overall health related quality of life and overall satisfaction with the treatment protocol were assessed using a five-point Likert scale. In addition, the following items were assessed using dichotomous (yes/no) scales as well as open-ended structured interview question format: benefits including changes to tongue range of motion, snoring, pain, sleep quality, nasal breathing (resolution of mouth breathing), speech, and swallow; and any complications or pain due to the surgery. For pre-pubertal children, the survey was completed by the parents. Continuous variables are summarized as mean (M) ± standard deviation (SD). Categorical variables are summarized as frequencies and percentages ± standard error (SE), where applicable.

Results

Our study included 348 patients with ages ranging from 29 months to 79 years. Demographic factors include age: 28.1 ± 20.2 years (M± SD); gender: 52.0% female; height: 146.2 ± 24.3 cm; weight: 57.1 ± 27.7 kg. This population includes 110 children (ages 2-11), 35 adolescents (age 12-17), 69 young adults (age 18-35), 120 adults (age 36-64), and 14 seniors (age ≥65). There were 63 children who were treated under general anesthesia in the operating room; all other cases were performed awake under local

anesthesia in the clinic. The average length of time from treatment date to follow-up was 4.3 ± 3 months, ranging from 2-20 months. Tongue tie severity (grades four through one, most to least severe, using the functional classification of ankyloglossia based on the tongue range of motion ratio) were graded as follows: 20.7% grade 4, 61.2% grade 3, 13.3% grade 2 with a posterior restriction, 4.7% grade 1 with a posterior restriction. Compensation patterns (floor of mouth elevation and muscular neck engagement to compensate for restrictive tongue mobility) that would affect the grading of tongue mobility were present and identified in 36.1% of cases. See Figure 2. There were 11.7% (n=41) of patients who had a prior frenectomy with persistent restrictions to tongue mobility prior to their participation in the current treatment. See Figure 3.

Benefits: There was an overall satisfaction rate of 91.1% (including 71.8% "very satisfied" and 19.3% "somewhat satisfied"), whereas 6.0% were neutral and 2.9% of patients reported dissatisfaction with the treatment protocol. See Table 1. Improvement to health-related quality of life was reported by 87.4%. See Table 2. Benefits reported by the patients included improvement to tongue mobility (96.5 ± 1.0 %); clenching or grinding of teeth (91.0 ± 4.3%); ability to perform myofunctional therapy exercises (89.8 ± 1.65%); ease of swallow (80.3 ± 3.5%); sleep quality (79.6 ± 2.6%); nasal breathing (78.4 ± 2.8%); neck, shoulder, facial tension or pain (77.5 ± 3.4%); and snoring (72.9 ± 3.4%). See Table 3.

Complications: There were 45.1% of patients who reported experiencing post-operative pain; average duration of pain was 3.3 ± 2.6 days with severity rating of 6.5 ± 1.9 (VAS: 0-10, mean ± SD). Severity of pain was most highly associated with depth of the surgical dissection and extent to which restrictions of the genioglossus muscle were released. Other factors associated with pain severity include: low tongue tone, less than ideal pre-operative myofunctional therapy compliance, prior myofascial pain syndromes, and patient declining to take post-operative pain medications.

Minor surgical site bleeding was reported by 12.6% of patients; the bleeding resolved in less than 3 hours among 58.9% of patients and 84.1% within 24 hours. There were 2.0% of patients that reported bleeding that lasted for more than 1 day. Numbness of the tongue-tip was reported by 4.86% of the patient population; numbness resolved within 2 weeks among 47.1% of patients, 70% within 2 months, and 99.7% within 6 months. There was only n=1 patient who reported tongue numbness beyond one year. Salivary gland issues were reported by 3.4% of patients; common issues included inflammation and swelling of the submandibular gland ducts, increased salivation, and jetting of saliva when lifting the tongue or eating. Most of these issues self-resolved within 1-2 weeks. See Table 4.

Other common issues include swelling, inflammation, mild wound infection, and potential scarring. Patients were recommended to rinse with salt water and/or alcohol-free mouthwash. Sutures usually fell out within 2-10 days. Gentle brushing of the wound after 5-7 days to debride granulation tissue with a soft surgical toothbrush (Curaprox CS Surgical Mega Soft) was found to be helpful. Premature wound opening was observed in a few cases and associated with sutures placed under high tension, failure to perform square knots when tying the sutures, patient protruding the tongue over the mandibular central incisors and tearing the stitches, and/or submucosal bleeding contributing to floor of mouth hematoma resulting in increased tension at floor of mouth. These wounds were left to heal by secondary intention and in some cases required revision surgery to excise scar tissue. There were 3.2% of patients who proceeded with a revision frenuloplasty procedure to excise scarring that resulted in worse mobility than prior to initial release; in addition, there were 3.4% of patients who elected to proceed with a second stage frenuloplasty to further improve tongue mobility after initial improvement. There were n=3 patients who had a third stage frenuloplasty; in these cases, the wound was left open to healing by secondary intention with good resolution as there was concern these patients may have had inflammation sensitivities to the suture material used for primary intention closure.

Two (n=2) patients reported worsened health symptoms after the procedure (0.6%) that were not associated with scar or wound healing issues; one of these patients was a patient with a narrow posterior airway space for whom measures of sleep-disordered breathing exacerbated after the procedure. See Figure 4. The other was a patient with narrow maxillary width and dental crowding treated for indication of mouth breathing who developed improved tongue resting posture and nasal breathing but reported pain from biting and clenching on the sides of the tongue with severe tongue scalloping due to insufficient tongue space. These patients were directed to maxillary and mandibular skeletal expansion as the next steps in their treatment.

Discussion:

Myofunctional therapy was first described in the medical literature by Alfred Paul Rogers in 1918 as an adjunct to orthodontic treatment to improve mandibular growth, nasal breathing, and facial appearance [16]. The foundational concepts he introduced regarding the importance of tongue-to-palate oral resting posture and nasal breathing for maxillofacial development were largely overlooked at that time despite a restatement of the myofunctional concept to the orthodontic community in 1950 [17]. Dr. John Mew, an English orthodontist, is credited for popularizing the Tropic Premise to his many disciples around the world [18, 19, 20] with the basic concept that the development of facial and dental structures is strongly influenced by the posture and function of the associated soft tissues (i.e., lips, tongue, orofacial

and mastication muscles) [19,21] and fortified by continuous nasal breathing [22 23-25]. Techniques for re-education of the orofacial muscles were published in French in the 1990s [26]. Even so, many thought leaders were slow to adopt these principles citing a lack of randomized control trials [27] and high-level evidence-based research [28]. Renewed interest for myofunctional therapy was garnered with a series of randomized control trials [29, 30,31]and cohort studies investigating the role of oropharyngeal exercises, speech therapy, myofascial re-education[32], and oro-nasal rehabilitation [33] for adults and children with sleep-disordered breathing. Furthermore, a more recent series of meta-analysis [12,13], review articles[34], books [35,36 2,37], commentaries [25,38], and position statements [39] have catapulted myofunctional therapy to the forefront of the attention within dental and medical communities, albeit not without criticism[40].

In this setting, there is increased attention to tongue-tie as a limiting factor for achieving one of the basic goals of myofunctional therapy: restoration or habituation of tongue posture to the roof of the mouth at rest (a.k.a., tongue-to-palate contact, lingual palatal suction). Restrictive lingual frenulum has been identified as a phenotype of obstructive sleep apnea in children [41 42] and adults [43], and recent studies on the assessment of functional ankyloglossia have been instrumental in identifying a larger population of patients with restricted tongue mobility [5].

A growing number of patients and providers are seeking peer-reviewed evidence-based information for the treatment of ankyloglossia, however, few investigators are publishing articles on this topic [44]. Most articles that are published on this topic consist of limited case-reports and case-series [45,46]; larger cohort studies are available on frenectomy techniques for infants as it relates to breastfeeding [47], however, there is still limited research relating to the release of tongue-ties among children [48], adolescents [49], and adults [50,51]. In this manuscript, we provide safety, efficacy, complication, and satisfaction results for the largest cohort of patients treated with lingual frenuloplasty and myofunctional therapy to date. Our results demonstrate a 91% patient satisfaction rate and an 87% rate of improvement in patient quality of life through a reduction in severity of mouth breathing, snoring, clenching, and/or myofascial tension. There was an overall minor complication rate of less than 5% (with no major complications reported to date) including risks of prolonged bleeding (2.0%), temporary numbness of the tongue-tip (4.9%), salivary gland issues (3.4%), and need for scar-excision revision surgery due to minor infection or poor wound healing (3.2%). In addition to improved tongue-mobility (96.5%) and an enhanced ability to perform myofunctional therapy exercises (89.8%), many patients in our cohort study expressed ameliorative effect in regard to clenching or grinding (91.0%), ease of swallow (80.3%), sleep quality (79.3%), nasal breathing (78.4%), release of neck, shoulder, or facial tension and pain (77.5%), and snoring (72.9%). The benefits attributed to improved oral function, breathing, and release of neck tension

are explained by resolution of oromyofascial dysfunction with potential mechanisms of action explored in a recent systematic review [34] and more completely explained in the peer-reviewed, evidence-based book *Tongue-Tied* by Baxter et al. [2].

The multidisciplinary treatment protocol combining frenuloplasty with myofunctional therapy as described herein was inspired and adapted from prior works [45, 46, 48, 52]. However, this cohort study is unique as all patients were required to demonstrate competence and compliance to myofunctional therapy for at least 1 month prior and 2 months after surgical treatment for tongue-tie. All procedures were performed with a scissors and suture technique without the use of laser or cautery. Moreover, the technique described in this manuscript involves release of submucosal genioglossus myofascial fibers (in addition to mucosal elements of the lingual frenulum), which may result in a more thorough release of submucosal restrictions but may also contribute to a greater severity of acute pain in the first 3-5 days immediately following the procedure. The application of sutures to close the wound after the release helps promote healing by primary intention to reduce the propensity for scar tissue and need for manual stretches.

Whereas many patients reported that the treatment protocol was "life-changing" with often dramatic patient testimonials available online (www.zaghimd.com), not all patients experienced similar outcomes. Indeed, many patients did not respond to treatment, and some expressed earnest dissatisfaction. Moreover, it should be emphasized that testimonials are not a scientific result and that long-term studies with objective findings are necessary to corroborate the findings of this preliminary report. Even so, the experience gathered from the patients who benefitted, as well as those who did not, has been enlightening and has allowed our team to develop the following guidelines for the release of tongue-tie in children and adults:

Guideline 1: Assessment of tongue-tie in children and adults requires evaluation of anterior tongue mobility based on tongue range of motion ratio, as well as an assessment of submucosal restrictions that may impair mobility of the posterior two-thirds body of the tongue.

Guideline 2: Whereas functional issues relating to tethered tongue mobility may linger over many years when first identified among adult patients, it is important to identify habituated compensatory patterns accommodating the restriction of tongue-tie in adult patients with impaired tongue mobility. Such compensation patterns may include engagement of the muscular neck, floor of mouth elevation, and lack of lingual-mandibular (tongue-jaw) disassociation with essential movements of the tongue.

Guideline 3: A comprehensive team for tongue-tie surgery requires an adequately trained surgeon as well as access to a supportive team including myofunctional therapist, physical therapist, craniosacral therapist, osteopathic specialists, and fascia specialists depending on the clinical circumstance.

Guideline 4: The goal of tongue-tie release in children, adolescents, and adults is to establish tongue tone, habituate correct posture, and enhance mobility; the tongue should remain in contact with the roof of the mouth at rest and normalization of a mature lingual-palatal swallow must be achieved at the completion of treatment. As such, pre and post-operative myofunctional therapy is essential for optimal preparation and recovery after tongue-tie surgery. The goals of pre-operative therapy are to create awareness of oral posture and tongue functions, improve tongue tone, and rehabilitate compensation patterns that may affect the post-operative recovery (e.g. floor of mouth elevation, muscular neck engagement, inability to perform isolated movements with the tongue without moving the jaw). For children, active parental involvement is critical in optimizing the success of the therapeutic program.

Guideline 5: Surgical release of the anterior tongue-tie is performed while the tongue is protruded up against the maxillary central incisors; release of posterior tongue-tie restrictions is performed while the tongue is engaged in lingual-palatal suction. This reinforces the need for pre-operative myofunctional therapy.

Guideline 6: We encourage non-traumatic release of lingual tissues that does not cauterize, burn, or injure surrounding or deeper structures. Whether the provider uses scissors or laser, it is critically important that only restrictive fibers are released and that excessive or indiscriminate use of cautery be avoided.

Guideline 7: Placement of simple-interrupted sutures using resorbable 4-0 or 3-0 chromic suture promotes healing by primary intention, the body's fastest and most efficient type of wound healing. If sutures are not used for primary intention closure or if the sutures fall out prematurely (sooner than 3-5 days), wound stretches are necessary to optimize healing by secondary intention to avoid wound scarring and contracture.

Guideline 8: Recommencing myofunctional therapy is essential for at least 2 months after a surgical release. The post-operative therapy focuses on optimizing wound healing as well as to re-educate tongue posture and optimal oral functions. Myofunctional therapy often continues for one year or longer (as needed) to prevent relapse of dysfunctional oral motor habits, promote exclusive nasal breathing, and ensure long-term habituation of ideal resting oral posture.

Guideline 9: Pre and post-operative photo documentation with the anterior tongue held up against the maxillary central incisors and with the tongue in lingual-palatal suction is recommended. Pre and post-operative documentation of tongue mobility and maximal incisal opening are also recommended.

Guidelines 10: Patients with limited tongue space in the maxilla and/or restricted posterior airway space are recommended to undergo a thorough evaluation of the structural determinants of the upper airway by a trained professional prior to tongue-tie release. Methods to assess posterior airway may include cone beam CT and/or flexible laryngoscopy. Lateral cephalogram is deemed insufficient for adequate assessment. Patients with posterior airway space less than 1 cm or maxillary dimensions limited for tongue space are recommended to consider dental orthopedic remodeling prior to tongue-tie release.

Providers of surgical interventions for tongue-tie release must understand that we as a community depend on each other to maintain a high quality of care for the betterment of our patients as well as for acceptance, standardization, and advancement of this field.

Conclusion: Lingual frenuloplasty with myofunctional therapy protocol as described in this manuscript is a safe and potentially effective treatment for mouth breathing, snoring, clenching, and cervico-facial tension in appropriately selected patient candidates. Further research will help to better identify the most optimal candidates for this treatment.

References:

1. Fernando C. Tongue Tie--from Confusion to Clarity: A Guide to the Diagnosis and Treatment of Ankyloglossia. Tandem Publications, 1998.
2. Baxter DM. Tongue-Tied: How a Tiny String Under the Tongue Impacts Nursing, Speech, Feeding, and More. Alabama Tongue-Tie Center, 2018.
3. Horton CE, Crawford HH, Adamson JE, Ashbell TS. Tongue-tie. Cleft Palate J 1969; 6.
4. Baxter R, Hughes L. Speech and Feeding Improvements in Children After Posterior Tongue-Tie Release: A Case Series. International Journal of Clinical Pediatrics 2018.
5. Yoon A, Zaghi S, Weitzman Ret al. Toward a functional definition of ankyloglossia: validating current grading scales for lingual frenulum length and tongue mobility in 1052 subjects. Sleep and Breathing 2017; 21:767-775.
6. Gross AM, Kellum GD, Hale STet al. Myofunctional and dentofacial relationships in second grade children. The Angle Orthodontist 1990; 60:247-253.
7. Yoon A, Zaghi S, Ha S, Law C, Guilleminault C, Liu S. Ankyloglossia as a risk factor for maxillary hypoplasia and soft palate elongation: A functional–morphological study. Orthodontics & craniofacial research 2017; 20:237-244.
8. Stecco C. Functional Atlas of the Human Fascial System E-Book. Elsevier Health Sciences, 2014.
9. Schultz RL, Feitis R. The endless web: fascial anatomy and physical reality. North atlantic books, 2013.
10. Scoppa F. Glosso-postural syndrome. Annali di Stomatologia 2005; 54:27-34.
11. Hanson ML. Oral myofunctional therapy. American journal of orthodontics 1978; 73:56-67.
12. Camacho M, Guilleminault C, Wei JMet al. Oropharyngeal and tongue exercises (myofunctional therapy) for snoring: a systematic review and meta-analysis. European Archives of Oto-Rhino-Laryngology 2018:1-7.
13. Camacho M, Certal V, Abdullatif Jet al. Myofunctional therapy to treat obstructive sleep apnea: a systematic review and meta-analysis. Sleep 2015; 38:669-675.
14. Moeller MR. The emerging area of orofacial myofunctional therapy: Efficacy of treatment in sleep disordered breathing bringing promise of a new field of medicine. CRANIO® 2018; 36:283-285.
15. Moeller JL, Paskay LC, Gelb ML. Myofunctional therapy: a novel treatment of pediatric sleep-disordered breathing. Sleep Medicine Clinics 2014; 9:235-243.
16. Rogers AP. Muscle training and its relation to orthodontia. International Journal of Orthodontia 1918; 4:555-577.
17. Rogers AP. A restatement of the myofunctional concept in orthodontics. American journal of orthodontics 1950; 36:845-855.
18. Mew J. The aetiology of malocclusion. Can the tropic premise assist our understanding? British dental journal 1981; 151:296.
19. Mew J. Tongue posture. British journal of Orthodontics 1981; 8:203-211.
20. Mew JR. The postural basis of malocclusion: a philosophical overview. American journal of orthodontics and dentofacial orthopedics 2004; 126:729-738.
21. Harvold EP, Chierici G, Vargervik K. Experiments on the development of dental malocclusions. American journal of orthodontics 1972; 61:38-44.
22. Harvold EP, Tomer BS, Vargervik K, Chierici G. Primate experiments on oral respiration. American journal of orthodontics 1981; 79:359-372.

23. McNamara Jr JA. Influence of respiratory pattern on craniofacial growth. The Angle Orthodontist 1981; 51:269-300.
24. Grippaudo C, Paolantonio E, Antonini G, Saulle R, La Torre G, Deli R. Association between oral habits, mouth breathing and malocclusion. Acta Otorhinolaryngologica Italica 2016; 36:386.
25. Torre C, Guilleminault C. Establishment of nasal breathing should be the ultimate goal to secure adequate craniofacial and airway development in children. Jornal de pediatria 2018; 94:101-103.
26. Chauvois A, Fournier M, Girardin F. Rééducation des fonctions dans la thérapeutique orthodontique: SID, 1991.
27. Mew J. Are random controlled trials appropriate for orthodontics? Evidence-Based Dentistry 2002; 3:35.
28. Mew JR. Does "evidence-based research" provide needed evidence? American Journal of Orthodontics and Dentofacial Orthopedics 2004; 125:A20.
29. Guimarães KC, Drager LF, Genta PR, Marcondes BF, Lorenzi-Filho G. Effects of oropharyngeal exercises on patients with moderate obstructive sleep apnea syndrome. American journal of respiratory and critical care medicine 2009; 179:962-966.
30. Diaferia G, Badke L, Santos-Silva R, Bommarito S, Tufik S, Bittencourt L. Effect of speech therapy as adjunct treatment to continuous positive airway pressure on the quality of life of patients with obstructive sleep apnea. Sleep medicine 2013; 14:628-635.
31. Villa MP, Brasili L, Ferretti Aet al. Oropharyngeal exercises to reduce symptoms of OSA after AT. Sleep and Breathing 2015; 19:281-289.
32. Guilleminault C, Huang Y, Monteyrol P, Sato R, Quo S, Lin C. Critical role of myofascial reeducation in pediatric sleep-disordered breathing. Sleep medicine 2013; 14:518-525.
33. Levrini L, Lorusso P, Caprioglio Aet al. Model of oronasal rehabilitation in children with obstructive sleep apnea syndrome undergoing rapid maxillary expansion: Research review. Sleep Science 2014; 7:225-233.
34. de Felício CM, da Silva Dias FV, Trawitzki LVV. Obstructive sleep apnea: focus on myofunctional therapy. Nature and science of sleep 2018; 10:271.
35. Gelb M, Hindin H. Gasp: Airway Health: The Hidden Path to Wellness. CreateSpace Independent Publishing Platform, 2016.
36. Liao F. Six-Foot Tiger, Three-Foot Cage: Take Charge of Your Health by Taking Charge of Your Mouth. Crescendo Publishing LLC, 2017.
37. Kahn S, Ehrlich PR. Jaws: The Story of a Hidden Epidemic. Stanford University Press, 2018.
38. Guilleminault C, Sullivan S. Towards restoration of continuous nasal breathing as the ultimate treatment goal in pediatric obstructive sleep apnea. Enliven: Pediatr Neonatol Biol 2014; 1:001.
39. Burger D. Pediatric airway symposium to clear the air on dental screening, treatment *ADA News*. https://www.ada.org/en/publications/ada-news/2018-archive/june/pediatric-airway-symposium-to-clear-the-air-on-dental-screening-treatment: American Dental Association, 2018.
40. Kezirian EJ. Oral myofunctional therapy and frenuloplasty are not proven treatments for obstructive sleep apnea. *Sleep-doctor.com (Dr. Kezirian's Blog)*. https://sleep-

doctor.com/blog/oral-myofunctional-therapy-and-frenuloplasty-are-not-proven-for-obstructive-sleep-apnea/, 2018.
41. Guilleminault C, Huseni S, Lo L. A frequent phenotype for paediatric sleep apnoea: short lingual frenulum. ERJ open research 2016; 2:00043-02016.
42. Huang Y, Quo S, Berkowski J, Guilleminault C. Short lingual frenulum and obstructive sleep apnea in children. Int J Pediatr Res 2015; 1.
43. Chien Y, Guilleminault C. Adult Short Lingual Frenulum and Obstructive Sleep Apnea. Journal of Sleep and Sleep Disorders Research 2017; 40:A164-A165.
44. Aaronson NL, Castaño JE, Simons JP, Jabbour N. Quality, Readability, and Trends for Websites on Ankyloglossia. Annals of Otology, Rhinology & Laryngology 2018:0003489418776343.
45. Tsaousoglou P, Topouzelis N, Vouros I, Sculean A. Diagnosis and treatment of ankyloglossia: A narrative review and a report of three cases. Quintessence international 2016; 47.
46. Junqueira MA, Cunha NNO, e SILVA Cet al. Surgical techniques for the treatment of ankyloglossia in children: a case series. Journal of Applied Oral Science 2014; 22:241-248.
47. Ghaheri BA, Cole M, Fausel SC, Chuop M, Mace JC. Breastfeeding improvement following tongue-tie and lip-tie release: A prospective cohort study. The Laryngoscope 2017; 127:1217-1223.
48. Ferrés-Amat E, Pastor-Vera T, Ferrés-Amat E, Mareque-Bueno J, Prats-Armengol J, Ferrés-Padró E. Multidisciplinary management of ankyloglossia in childhood. Treatment of 101 cases. A protocol. Medicina oral, patologia oral y cirugia bucal 2016; 21:e39.
49. Lalakea ML, Messner AH. Ankyloglossia: the adolescent and adult perspective. Otolaryngology—Head and Neck Surgery 2003; 128:746-752.
50. Lalakea ML, Messner AH. Ankyloglossia: does it matter? Pediatric Clinics of North America 2003; 50:381-397.
51. Chinnadurai S, Francis DO, Epstein RA, Morad A, Kohanim S, McPheeters M. Treatment of ankyloglossia for reasons other than breastfeeding: a systematic review. Pediatrics 2015:peds. 2015-0660.
52. Fabbie P, Kundel L, Vitruk P. Tongue-Tie Functional Release. Dent Sleep Practice Winter 2016:40-45.

Figure Legends

Figure 1. Case example: 19-year-old man presenting with mumbling, drooling, unrefreshing sleep, fragmented sleep, and chronic mouth breathing associated with Grade 3 tongue-tie (<50% mobility of the tongue-tip to the incisive papilla compared to maximal incisal opening). Note the compensation patterns of floor of mouth elevation and tension on the attached gingiva due to the restrictive lingual frenulum. Baseline images obtained after preparation with pre-operative myofunctional therapy, immediately prior to surgical release. Immediate post-op images show excision of the mucosal frenulum and submucosal myofascial fibers with primary intention closure using 4-0 chromic suture. Note the release of tension from the floor of mouth and attached gingiva, as well as the improved tongue mobility. Photos are taken in neutral position, tongue elevated to the central incisors, and while in suction-hold (i.e. lingual-palatal suction, "cave".)

Figure 2. Case Example: 6-year-old girl with restless sleep, nail biting, dental grinding, and open mouth breathing presenting with Grade 3 compensating to Grade 2 tongue-mobility. The image on the left shows <50% mobility (Grade 3 TRMR) with floor of mouth elevation and tension on attached gingiva. The image on the right shows 50-80% mobility (Grade 2), however, the patient exerts extensive strain from the floor of mouth and muscular neck to compensate for the restricted tongue mobility.

Figure 3. Case example: 16-year-old boy with Grade 4 tongue-tie (<25% TRMR) with persistently restricted tongue mobility (Grade 3, <50% TRMR) despite initial laser frenectomy (performed elsewhere) who was rehabilitated to Grade 1 mobility (>80% TRMR) with lingual frenuloplasty and myofunctional therapy protocol.

Figure 4. Use of computed tomography imaging to assess for tongue-space in the assessment of candidates for lingual frenuloplasty. The midline, sagittal image reconstruction of the CT scan is used to assess the available space for the tongue in the oral cavity. Note that despite both patients having similarly restricted amount of posterior airway space, the patient on left has no space between the tongue and the palate (poor candidate), while the patient on the right has a significant amount of space between the tongue and the palate (better candidate). Lingual frenuloplasty and myofunctional therapy is considered to be less effective in patients without adequate oral volume for tongue-space. Such patients may be better suited to dental orthopedic remodeling (orthodontics and/or orthognathic surgery for expansion and advancement of the skeletal framework) to increase the tongue-space in addition or prior to treatment with lingual frenuloplasty.

Table 1. Patient-reported satisfaction with lingual frenuloplasty and myofunctional therapy treatment protocol.

Satisfaction	Number	Percent Total	
A (very satisfied)	250	71.8%	**Overall Satisfied: 91.1%**
B (somewhat satisfied)	67	19.3%	
C (neutral)	21	6.0%	
D (somewhat dissatisfied)	10	2.9%	**Overall Dissatisfied: 2.9%**
F (very dissatisfied)	0	0.0%	

Table 2. Health-related quality of life following lingual frenuloplasty and myofunctional therapy treatment protocol.

Health-Related Quality of Life	Number	Percent Total	
A (much better)	137	39.3%	**Overall QOL Improved: 87.4%**
B (somewhat better)	167	48.0%	
C (neutral)	42	12.1%	
D (somewhat worse)	2	0.6%	**Overall QOL Worse: 0.6%**
F (much worse)	0	0.0%	

Table 3. Benefits attributed to lingual frenuloplasty with myofunctional therapy protocol.

Benefits	Improved	Did Not Improve	Unsure	N/A	Percent Improved	Standard Error
Overall tongue mobility	326	12	10	-	**96.5%**	1.0%
Clenching or grinding of teeth	40	4	-	304	**91.0%**	4.3%
Ability to perform myofunctional therapy exercises	307	35	6	-	**89.8%**	1.6%
Ease of swallow	102	25	3	218	**80.3%**	3.5%
Sleep quality	195	50	11	92	**79.6%**	2.6%
Nasal breathing	174	48	4	122	**78.4%**	2.8%
Neck, shoulder, facial tension or pain	117	34	-	197	**77.5%**	3.4%
Snoring	102	38	11	197	**72.9%**	3.8%

Table 4. Patient reported risks and complications associated with lingual frenuloplasty.

Risks/ Complications	Reported	Not Reported	**Percent Reported**	Standard Error
Pain	157	191	**45.1%**	2.7%
--- Pain for longer than 7 days	5	343	**1.4%**	0.6%
Bleeding	44	304	**12.6%**	1.8%
--- Prolonged bleeding >24 hours	7	341	**2.0%**	0.8%
Numbness of the tongue-tip	17	331	**4.9%**	1.2%
--- Numbness >2 weeks	9	339	**2.6%**	0.9%
Salivary gland issues	12	336	**3.4%**	1.0%
--- Complaints> 2 weeks	3	345	**0.9%**	0.5%
Second stage release procedure to further improve tongue mobility after initial improvement	12	336	**3.4%**	1.0%
Revision surgery to excise scarring that resulted in worse mobility than prior to initial release	11	337	**3.2%**	0.9%

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203x182mm (96 x 96 DPI)

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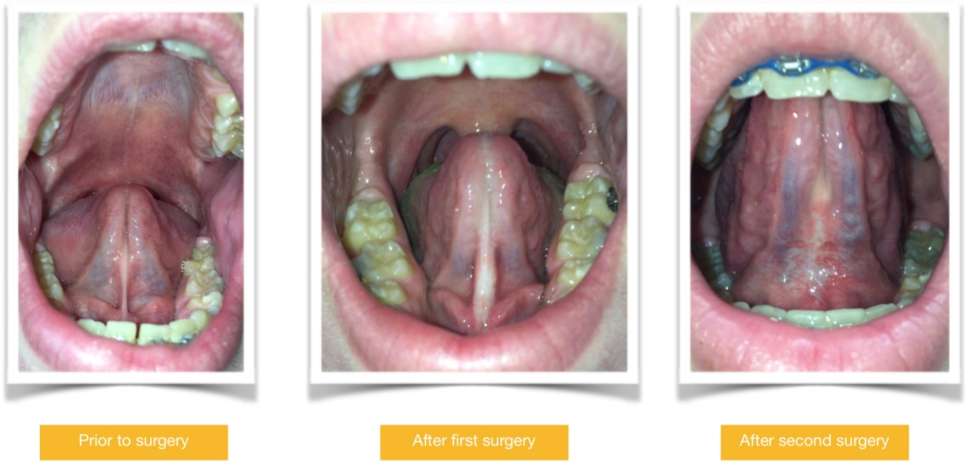
204x101mm (144 x 144 DPI)



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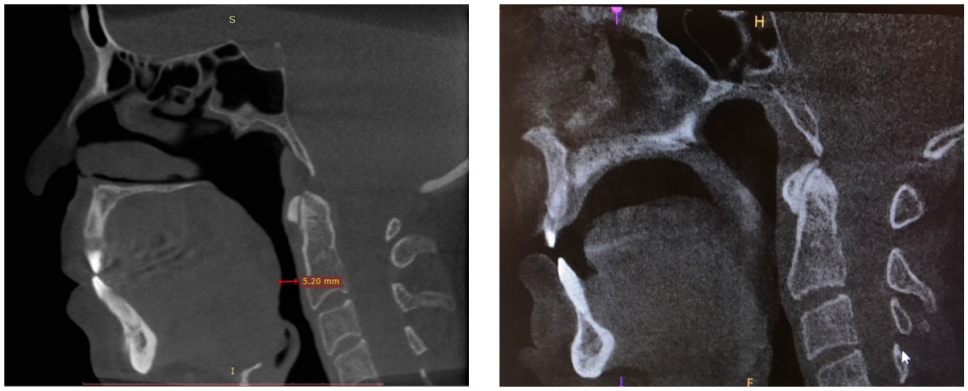

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219x92mm (144 x 144 DPI)