Ankyloglossia in Children and those with Special Healthcare Needs: Diagnosis Management and Implications

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ABSTRACT

Oral ties along with ankyloglossia have been recognized for centuries, but have gained wider interest in the recent years. Tongue-tie refers to a congenital condition characterized by a short lingual frenulum that restricts tongue movement and interferes with tongue function. Recent literature suggests that speech, feeding, oral hygiene, dentition, and sleep difficulties may be linked to this condition. This case series presents evidence on the diagnosis, management using soft tissue laser with the help of pharmacological/nonpharmacological behavior management techniques, use of myofunctional exercises and future implications of short lingual frenulum in young children.

Keywords: Ankyloglossia, Feeding difficulty, Frenectomy, Soft tissue laser, Speech difficulty, Tongue-tie.

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INTRODUCTION

"Ankyloglossia, or tongue-tie, is a congenital anomaly that is characterized by a short lingual frenulum."¹ By definition, it represents a condition in which the tongue's movement is restricted due to the lingual frenulum "tethering" the tongue, affecting speech clarity and hence the name "tongue-tie." Anatomically the tongue fascia located beneath the oral mucosa is fused with the connective tissue and extends down to diaphragm into the hip the way down to arches of the feet. These fascial layer surrounds sublingual glands and submandibular ducts and anterior genioglossus fiber are located beneath it. Superficially lingual nerve branches seen on the ventral surface of tongue, immediately deep to fascia.²

Worldwide, the incidence of ankyloglossia ranges between 4 and 10.7% in different populations with a slight male predilection.³ Prevalence in Indian population has been reported to be as low as 0.02–0.4%.⁴ The low prevalence could be on account of lack of standardized protocols among health-care practitioners for its early identification and low awareness among the population in general. Both environmental and genetic factors are related to its etiology. Molecular analysis found that point mutations with *TBX22* gene located on Xq21 can lead to cleft palate and ankyloglossia. Epidermolysis bullosa, specifically recessive dystrophic subtype has been associated with ankyloglossia. Other associated syndromes include Opitz, orofacial digital, Beckwith-Weidemann, Simpson-Golabi-Behmel, Van de Woude, and Pierre Robin.⁵

Functionally the most important quality of tongue is to elevate rather than protrusion for feeding, nursing, speech, and dental arch development. Historically, tongue-tie has been associated mainly with speech difficulties and thus more emphasis has always been given to its functional, rather than an anatomical diagnosis. At the time of development of speech around age of 2–3 years patients having tongue-tie usually have difficulty in articulation of speech and sounds like /s /, /z /, /t /, /d /, /l /, /sh /, /ch /, /th /, and /dg /, particularly difficulty to roll an R.⁶

The recent literature shows emerging concerns about its association with different direction of growth of maxilla and

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mandible, with reduced airflow leading to breathing problems, orthodontic problems, and altered habits such as mouth breathing, forward positioning of the tongue. There is also a evidence, though limited, which shows abnormal tongue position or ankyloglossia can lead to Class III malocclusion.⁷ As tongue is prevented from resting on the palate due to tongue-tie which can also lead to problem like sleep difficulties and sleep apnea in children and adults. This lack of tongue to palate suction, during sleep, allows the tongue to fall into the pharynx restrict or obstruct the airway and leads to SBD or sleep apnea.^{8,9} The associated problems due to ankyloglossia in different age groups are listed in (Table 1).

Clinically, to grade the extent of ankyloglossia, Kotlow's classification has most commonly used which is based on the length of tongue from the insertion of lingual frenum into base to the tip of tongue; "Class I, mild ankyloglossia 12–16 mm; Class II, moderate ankyloglossia 8–11 mm; Class III, severe ankyloglossia 3–7 mm; and Class IV, complete ankyloglossia <3 mm. In a tongue with normal function and range of movement, interincisal distance by maximal mouth opening, while maintaining contact of the tongue-tip to the posterior surface of the upper central incisor teeth should be >30 mm."¹⁰

Another, more recent grading scale for functional classification of ankyloglossia is based upon tongue range of motion ratio (TRMR), expressed as percentage of ratio of maximum interincisal distance when tongue tip lies on Incisal Papilla (MOTTIP) to maximal interincisal mouth opening (MIO) as: "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.¹¹

There have been studies reporting differences among pediatricians, dental specialist's lactation consultants, otolaryngologists, speech pathologists, and surgeons in their treatment recommendations. A lack of consensus regarding accepted anatomical and diagnostic criteria for degree of restriction and impact on growth, development, feeding, or oral motor function leads to controversy regarding clear indications and timing of surgical treatment.⁶ Keeping in view, the recent literature, early intervention for ankyloglossia may prove to be more beneficial for the growing child. The pain-free treatment modalities such as soft tissue lasers further make it easier for the clinicians to deliver a pain-free experience to the young children. The growing awareness among parents and greater concern for the well-being of their children is also leading to a trend of early reporting for seeking treatment and thus a greater need for the clinicians to generate more evidence-based guidelines for frenectomies/frenotomies.

This case series compiles frenectomies done in patients with pathological lingual frenulum, and impaired functions having reported with different medical/behavioral background.

Case 1

A 7-year-old male child presented to the Pediatric Dentistry Unit, Oral Health Sciences Centre, PGIMER with a complaint of "pain in upper right back tooth region." The patient had no significant

medical history. Tongue-tie was noticed as an incidental finding (Figs 1A to C) with a suspicion due to lisping noticed in child by the pediatric dentist while communicating with him at the time of examination. Parents reported that child had difficulty in pronouncing the sibilants and had not received any previous medical intervention, which highlights the poor awareness about the condition. Clinical intraoral examination showed difficulty in exercising tongue mobility (TRMR of 8.9%; Grade 4) due to short lingual frenula and a deep carious lesion in the maxillary right second primary molar. Since the child was cooperative, his chief complaint was first addressed and laser-assisted lingual frenectomy was later performed, after taking parental consent (Fig. 1D). Bilateral lingual nerve block was given to reduce postoperative discomfort and diode laser (Biolase, at power 1 W, CP 2 mode) was used taking adequate precautions by the operator and the patient in a separate operatory. No suturing was done and patient was advised to maintain a soft diet, regular oral hygiene, and take analgesics as needed. He was recalled after 1-week and 1-month. Tongue movement was checked by protrusion to assess complete elimination of the frenum and lingual mobility at 1-week postop showed an improvement with interincisal distance while tongue touching the palate to be 15 mm. Myotherapy exercises were advised to the patient.¹² At 1-month follow-up complete healing along with improvement in speech was observed (Figs 1E and F). The patient was advised to undergo speech therapy for correction and improvement of their speech.

Table 1: Impact of ankyloglossia at various age-groups^[19,20]

	Infancy	Toddler	Children	Adult
Impact of tongue-tie	Bottle and breastfeeding challenges	Eating challenges, malocclusion, poor oral health	Postural issues	
	Poor tongue function, mouth breathing, airway obstruction, and snoring, aberrant swallowing, obstructive sleep apnea

Figs 1A to F: (A–C) Preoperative; (D) Intraoperative; (E–F) 1-month follow-up postoperative

Case 2

A 12-year-old female child presented to our unit with a complaint of difficulty in pronouncing certain words. The child was shy to speak at school and felt embarrassed due to her inability to pronounce certain words with "t" and "d" sound properly. The parents had consulted a speech pathologist and had been advised to seek an opinion for the apparently short lingual frenum. There was no significant medical history. Intraoral examination revealed a restricted tongue mobility due to a short lingual frenum assessed to be grade 4 (TRMR- 6.6%), (Fig. 2A). Laser-assisted lingual frenectomy was performed, after parental consent (Fig. 2B). One-week postop showed an improvement with interincisal distance while tongue touching the palate to be 13 mm (Fig. 2C). Myotherapy exercises were advised along with the speech therapist consultation.

Case 3

A 4-year-old male child was referred to our unit from Advance Paediatric Centre with a complaint of difficulty in speech. Patient was a known case of autism spectrum disorder and mixed cerebral palsy. Patient had a history of seizures and high-grade fever 10 days after birth and delayed developmental milestones. He had no history of seizures after the first episode and at the time of examination was not on any antiepileptic medications. Intraoral examination revealed short lingual frenulum with limitation of tongue function (Fig. 3A). As the child was uncooperative, laser-assisted lingual frenectomy was performed under general anesthesia (Figs. 3B and C). The procedure was successfully completed and the recovery was uneventful. At follow-up visits the parents reported an improvement in the overall well-being of the child due to a significant adequate food intake. The improvement in speech, however, could not be assessed in the short span due to the child's medical condition.

Case 4

A 3-year-old male child was brought by his parents complaining of difficulty in feeding and speech in patient. There was no significant medical history. Intraoral examination revealed short lingual frenula with limitation of tongue function. As a child was very young a laser-assisted lingual frenectomy was performed under general anesthesia. The procedure was successful and recovery was uneventful. At postoperative follow-up at 1-month, parents reported an improvement in diet as well as the child's vocabulary to 15 words from just 2–3 words preoperatively.

DISCUSSION

The tongue plays a vital role in skeletal and oro-facial development. It maintains a balance of forces between growing maxillofacial skeleton and soft tissue structures. The pathological lingual frenulum, can lead to problems that can be solved only and exclusively with multi-disciplinary approaches which involves a pediatrician, pediatric dentist, Otolaryngologist, lactation consultants, craniosacral therapist, sleep consultants, and speech therapist for the best clinical outcomes for the child, allowing normal physiologic growth and development. The pediatric dentist plays a key role in the diagnosis and treatment by intervening therapeutically, evaluating the growth and development of cranium-facial area, breathing, dental malocclusion, mobility, and posture of lingual frenulum.

Many treatment modalities are available for ankyloglossia which include conventional surgical techniques, electrocautery, and lasers for frenotomy as well as frenectomy.¹³ Laser technology, as used in the present cases and also reported in the literature, with fewer postoperative complications (swelling and infection), capillaries are sealed by protein denaturation and clotting factor VII is stimulated. Additional advantages include a shorter operating time, improved ability to control bleeding, and fewer intra and postoperative pain and discomfort.¹⁴ A review by Garrocho-Rangel stated that use

Figs 2A to C: (A) Preoperative; (B) Immediate postoperative and, (C) One-week postoperative

Figs 3A to C: (A) Preoperative; (B) Intraoperative laser assisted procedure; (C) Immediate postoperative

Table 2: Postoperative myofunctional exercises^[13]

Exercises	Advantages
<ul style="list-style-type: none"> Repeat stretching the tongue up toward the nose and down toward the chin With the mouth wide open, place the tongue on the front teeth and touch them Close your mouth and press on your left and right cheeks to form a lump. 	<ul style="list-style-type: none"> Develops new muscle movements, involving tongue-tip Increases kinesthetic awareness of full range of movements of tongue Encourages movements like licking of lips, cleaning of anterior and posterior teeth.

of lasers for ankyloglossia for children under 15 years of age is more preferable compared to standard techniques.¹⁵ According to histological studies, laser wounds contain significantly fewer myofibroblasts, which means less wound contraction and scarring, and ultimately improved healing over scalpel techniques.^{16–18}

All the cases treated in present series showed minimal bleeding intraoperatively with no need for suture placement, minimal postoperative discomfort and uneventful healing.

Apart from the routine postoperative care, an important factor that needs to be emphasized to the parents, is the tongue exercises advised to the patients, which determine the overall long-term positive outcome of the surgical procedure. The patients in the present series were advised tongue exercises (Table 2) every day for a period of 3–4 weeks, for 3–5 minutes at a time. The Cases 1 and 2 showed good compliance with the exercises as their TRMR showed significant improvement. The Cases 3 and 4 of the present series showed questionable compliance with these exercises on account of behavioral issues but showed significant improvement functionally. Overall the parental satisfaction with the treatment rendered in all cases was good.

CONCLUSION

A pediatrician or a pediatric dentist should always do a systematic assessment for the children with tongue-tie and direct timely referral and treatment when functions are impaired. A reliable and compressive guideline to classify, diagnose, treat, and measure outcome parameters should be formulated taking into account all the disciplines involved with these cases which will enable clinicians to effectively communicate about the level of oral and nasal functional impairment due to limited tongue mobility which in turn affects facial development.

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