ORIGINAL RESEARCH

Sequelae of Tethered Oral Tissues in Infants: A Challenging Expertise Conundrum

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Abstract

Timely prediction of diversified effects of tethered oral tissues in infancy is utmost important for the proper development of the stomatognathic system.

Objective: To assess current opinion/knowledge of infant oral care specialists about the repercussions of tethered oral tissues in infants influencing function, growth, and development.

Study design: A cross-sectional survey was done among 192 dentists of which 46.35% were oral surgeons and 53.65% were pediatric dentists. The responses obtained were subjected to the statistical analysis using the Chi-square test.

Results: Most commonly reported conjectures include speech defects (77.6%), breastfeeding dysfunction (71.8%), midline spacing between teeth (71.4%), atypical swallowing (67.7%), followed by sleep issues (31.8%) and dentoskeletal alterations (43.2%). Least possibly reported conjectures are postural alterations (10.4%), caries susceptibility (13.5%), unexpected and unexplained asphyxia (15.6%), and tearing of gingival tissues (19.3%).

Conclusion: Both specialty dentists, i.e., pediatric dentists and oral surgeons, believe that tethered oral tissues cause breastfeeding dysfunction, speech impediments, midline diastema, and permanence of atypical swallowing but limited awareness exists about their consequences such as sleep and breathing disorders, caries initiation, gingival recession, malocclusions, and postural alterations.

Keywords: Infants, Lip tie, Tethered oral tissues, Tongue tie.

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Introduction

The oral frenulum is a thin fibrodense conjunctive fold of mucous membrane enclosing muscle fibers. It extends as a cord-like tissue connecting the lip, buccal mucosa, or tongue to the alveolar mucosa.[1] During embryonic development, these thin membranes shorten and migrate due to programmed cell death.[2] In some cases, an erroneous cellular death process might lead to tethered oral tissue, notably tongue tie and lip tie, and is a congenital anomaly that limits the normal functioning of the stomatognathic system. Tethered oral tissues influence the child's growth and development that exists in a continuum with a broad clinical spectrum from inapparent to severe and fulminating episodes and their diagnosis requires in-depth knowledge. Pediatric dentists and oral and maxillofacial surgeons are mostly commonly involved specialists who interact with infants in dentistry.[3] The present study was designed to assess opinion/knowledge of specialty dentists such as oral and maxillofacial surgeons (OMFSs) and pediatric dentists in present days concerning the effects of tethered oral tissues on functions like breastfeeding, sleep, etc., and their further impact on growth and development.

Materials and Methods

Between June and August 2020, a cross-sectional, population-based, questionnaire study was conducted following the STROBE guidelines.

Questionnaire Validation

A self-administered questionnaire was framed after thorough review of the literature. Questions were both closed-ended and open-ended. The queries regarding the demographic information were included in the first section of the questionnaire. The second section had 10 questions regarding the probable influence of tethered tissues on the function and development of the infant with a three-point Likert scale (yes, no, maybe). Total 20 specialty dentists were selected and a printed questionnaire was given to pretest the validity of the questionnaire and determine the feasibility of the study. These duly filled responses were not included in the final analysis. The validity of the questionnaire was checked by the test-retest method after 15 days. A Cronbach's alpha value of 0.82 was obtained, which showed good internal reliability of the questionnaire.

Inclusion Criteria

Pediatric dentists or oral maxillofacial surgeons with at least 1 year of clinical experience

Exclusion Criteria

(1) The dentist who refused to participate in the study. (2) Postgraduate students in pediatric dentistry. (3) Postgraduate students in oral and maxillofacial surgery.

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Study Design

The study was commenced after obtaining approval from the institutional ethical committee and review board. As this was a population-based study, no sample was drawn. A cross-sectional survey was undertaken among the infant oral care professionals like pediatric dentists and oral maxillofacial surgeons using snowball sampling—a nonprobability sampling method. It relies on referrals from initial respondents to generate additional respondents. The initial respondents here include the faculty of different dental colleges in Andhra Pradesh. Only one referral from each respondent was chosen and this progressed in a chain reaction. Respondents' names were not elicited in the questionnaire in order to enhance participation and to maintain confidentiality.

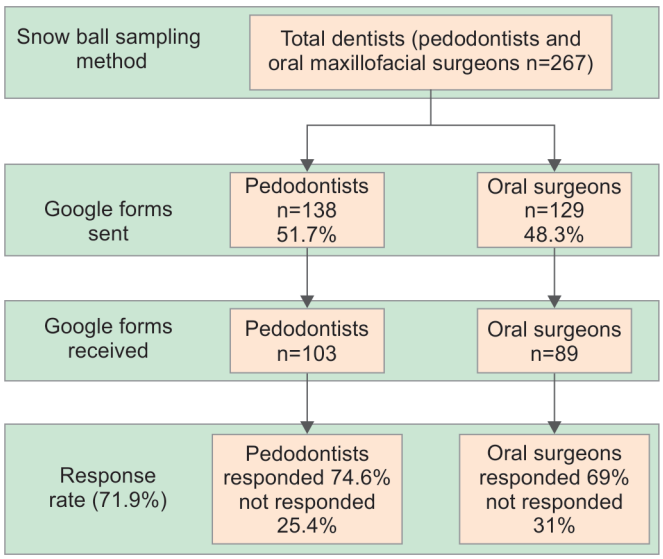
Google forms containing self-administered, open- and closed-ended questionnaire were sent through e-mail to almost 267 specialty dentists. Remainder mails were sent after 1 week and telephone calls were made to the nonresponders in an attempt to maximize the response rate. Responses obtained were tabulated and statistically analyzed using the Pearson's chi-square test.

Results

A response rate of 71.9% (n = 192) was obtained, out of which 53.65% (n = 103) were pediatric dentists and 46.35% (n = 89) were oral maxillofacial surgeons with roughly balanced proportion (Flowchart 1). Table 1 shows the study population demographics. Total 123 (64.1%) males and 69 (35.9%) females with mean age of 34.93 years have participated in the survey. About 51% participants had <5 years' experience and 49% had >5 years' experience within a range of 3.5–28 years of experience.

Figure 1 represents the graphical representation of the relative opinion/knowledge of both specialty dentists about the impact of tethered tissues (tongue tie/lip tie) on infant's oral cavity and further development. When questioned its effects on breastfeeding, majority of target population (69.9% pediatric dentists; 74.22% surgeons) are in opinion that tethered tissues can cause breastfeeding dysfunction and, statistically, there is no significant difference in the opinion between the specialties (p = 0.794) and 24.3% pediatric dentists; 21.3% surgeons were doubtful about the association. When enquired regarding the after effects of tied oral tissues on the child development, the most possible complications believed to be associated are articulation issues and speech delay (84.3% pediatric dentists; 69.7% surgeons), midline spacing between teeth (72.8% pediatric dentists; 69.7% surgeons), and atypical swallowing (71.8% pediatric dentists; 62.9% surgeons). Few dentists are in opinion that the tethered tissues are positively associated with sleep disordered mouth breathing (31.8%) and morphological dentoskeletal alterations (43.2%). Least response was obtained regarding the positive association of tethered tissues with postural alterations (10.4%), caries susceptibility (13.5%), unexpected and unexplained asphyxia in infancy (15.6%), and pathological tearing of gingival tissues (19.3%) as shown in Figure 2 and Table 2.

Flowchart 1: Sample distribution



Discussion

The Dental Home concept recommends first dental visit within 6–12 months of age to reduce the child's risk of oral diseases.[4] It is essential that the dental specialists who perform elective surgical procedures in infants on a daily basis should be familiar with all possible pathologies occurring during this early phase of life.[5]

Tethered oral tissues are congenital anomalies that include short and/or thick, frenulum that ties and limits the movement of either tongue, lips, or cheeks.[3] These tethered tissues can disrupt the harmonious balance of the stomatognathic system resulting in wide spectrum of consequences with functional limitations during infant's growth and subsequently in child's development. Early diagnosis and intervention of tethered oral tissues in infancy requires comprehensive knowledge of their impact on child's development. The current study has revealed differing views of related specialty dentists regarding the sequelae of tethered oral tissues.

Expertise Quandary in Spectrum of Consequences of Tethered Oral Tissues

The present study showed that the target population had a better perspective about the influence of tethered oral tissues on breastfeeding with a satisfactory result (71.8%). About 22.9% of the total sample were not sure about the ill effects of tongue tie or lip tie on breastfeeding mechanism. There is no significant difference observed between opinion of pediatric dentists and oral surgeon ($p > 0.05$).

Table 1: Sample demographics

Variable		*Pediatric dentists N = 103 (53.65%)*	*Oral maxillofacial surgeon N = 89 (46.35%)*	*Total N = 192 (100%)*
Age	<35 years	69 (67%)	49 (55.1%)	118 (61.5%)
	>35 years	34 (33%)	40 (44.9%)	74 (38.5%)
Gender	Male	53 (51.5%)	70 (78.7%)	123 (64.1%)
	Female	50 (48.5%)	19 (21.3%)	69 (35.9%)
Clinical experience	0–5 years	55 (53.4%)	43 (48.3%)	98 (51%)
	6–10 years	23 (22.3%)	16 (18%)	39 (20.3%)
	11–15 years	10 (9.7%)	16 (18%)	26 (13.5%)
	>15 years	15 (14.6%)	14 (15.7%)	29 (15.1%)

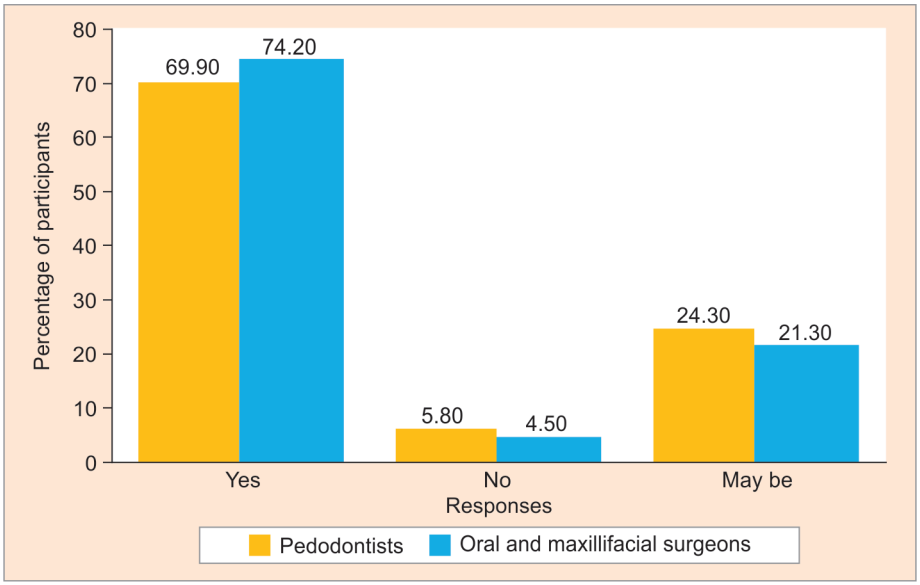


Fig. 1: Opinion regarding breastfeeding in infancy

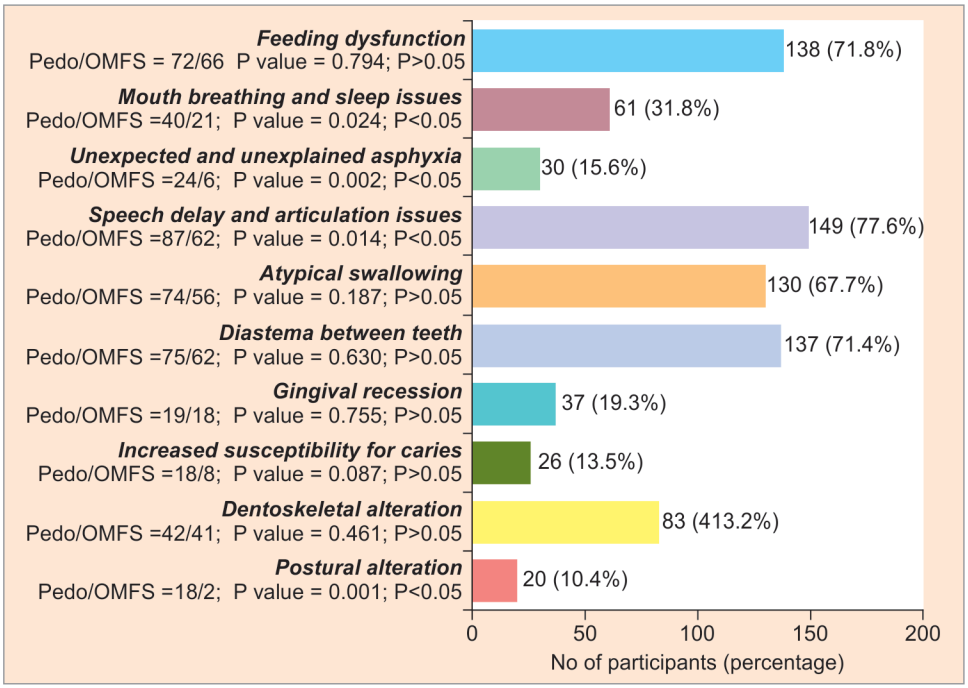


Fig. 2: Pediatric dentist's and oral surgeon's perception about probable sequel of tethered tissues, which influences the function, growth, and development from infancy through adolescence

The suck-swallow-breath mechanism in infants utilizes lips, flanging around the breast, to create a seal for sucking and dorsum of the tongue to latch onto the nipple and actively squeeze milk from the breast into the oral cavity for swallowing.[6] This mechanism is more difficult for infants with tongue tie and/or upper lip ties causing breastfeeding issues such as painful unsustained latch,[3] aerophagia,[7] colic reflux,[8] failure to thrive,[9,10] etc., in infant and sore nipples, trauma, mastitis, etc., in mothers.[3,5,11] Some tethered tissues like anterior tongue tie/lip tie are highly visible and easier to detect. However, posterior tongue ties are more challenging to diagnose.[12] In a prospective study, 91% mothers have reported an overall improvement in breastfeeding after the frenotomy procedure and also infants' ability to extend their tongues to both the lower gum and the lower lip was significantly increased post-frenotomy compared to preprocedure.[13]

Only 31.8% specialists perceive that the severe form of tongue tie/lip tie results in open mouth posture, which in turn develops mouth breathing subsequently sleep disturbances with significant difference in the opinions among pediatric dentists and oral surgeons ($p < 0.05$).

Tongue ties and low tongue resting postures often lead to or exacerbate mouth breathing and cause abnormal development

Table 2: Pediatric dentists and oral maxillofacial surgeons' aptitude toward spectrum of consequences due to tethered oral tissues

Association of tethered oral tissues	*Target population*	*Yes n (%)*	*No n (%)*	*Maybe n (%)*	*p value*
Q1. Does a tethered oral tissue (lip tie/tongue tie) cause breastfeeding problems in infancy?	Pediatric dentists	72 (69.9)	6 (5.8)	25 (24.3)	0.794
	Oral surgeons	66 (74.2)	4 (4.5)	19 (21.3)	
	Total	138 (71.8)	10 (5.2)	44 (22.9)	
Q2. Does lip tie/tongue tie cause sleep disturbance due to mouth breathing in infancy?	Pediatric dentists	40 (38.8)	63 (61.2)	0 (0)	0.024*
	Oral surgeons	21 (23.6)	68 (76.4)	0 (0)	
	Total	61 (31.8)	131 (68.2)	0 (0)	
Q3. Is there any chance for unexpected and unexplained asphyxia in infants due to tethered oral tissues?	Pediatric dentists	24 (23.3)	79 (76.7)	0 (0)	0.002**
	Oral surgeons	6 (6.7)	83 (93.3)	0 (0)	
	Total	30 (15.6)	162 (84.4)	0 (0)	
Q4. Does tongue tie in infancy cause articulation issues and speech delay?	Pediatric dentists	87 (84.5)	16 (15.5)	0 (0)	0.014*
	Oral surgeons	62 (69.7)	27 (30.3)	0 (0)	
	Total	149 (77.6)	43 (22.4)	0 (0)	
Q5. Does a tethered oral tissue in infancy lead to development of atypical swallowing?	Pediatric dentists	74 (71.8)	29 (28.2)	0 (0)	0.187
	Oral surgeons	56 (62.9)	33 (37.1)	0 (0)	
	Total	130 (67.7)	62 (32.3)	0 (0)	
Q6. Does a severely tethered oral tissue in infancy cause midline diastema during development of dentition?	Pediatric dentists	75 (72.8)	28 (27.2)	0 (0)	0.630
	Oral surgeons	62 (69.7)	27 (30.3)	0 (0)	
	Total	137 (71.4)	55 (28.6)	0 (0)	
Q7. Does a severely tethered oral tissue in infancy cause retraction of gingiva tissue?	Pediatric dentists	19 (18.4)	84 (81.6)	0 (0)	0.755
	Oral surgeons	18 (20.2)	71 (79.8)	0 (0)	
	Total	37 (19.3)	155 (80.7)	0 (0)	
Q8. Does severely tethered oral tissue in infancy increase the susceptibility to caries?	Pediatric dentists	18 (17.5)	85 (82.5)	0 (0)	0.087
	Oral surgeons	8 (9.0)	81 (91.0)	0 (0)	
	Total	26 (13.5)	166 (86.5)	0 (0)	
Q9. Does a severely tethered oral tissue in infancy lead to the development of morphological dentoskeletal malocclusion?	Pediatric dentists	42 (40.8)	61 (59.2)	0 (0)	0.461
	Oral surgeons	41(46.1)	48 (53.9)	0 (0)	
	Total	83 (43.2)	109 (56.8)	0 (0)	
Q10. Does a severely tethered oral tissue in infancy can lead to postural alterations?	Pediatric dentists	18 (17.5)	85 (82.5)	0 (0)	0.001**
	Oral surgeons	2 (2.2)	87 (97.8)	0 (0)	
	Total	20 (10.4)	172 (89.6)	0 (0)	

*$p < 0.05$—significant; **$p < 0.005$—highly significant

of the oral cavity, increasing the risk of upper airway collapse during sleep.[9,14] The American Academy of Pediatrics has advised for screening tongue positioning as it contributes to airway obstruction and sleep disordered breathing.[15] About 64% of brain growth occurs in the first 90 days after birth; it is considered as critical time for the developing brain. Disturbed sleep reduces the production of growth hormone, which subsequently leads to retarded growth.

Neurodevelopmental complications of inadequate sleep include developmental delay, poor school performance, hyperactivity, aggressive behavior, and social withdrawal.[9] Signs and symptoms like snoring, upper airway resistance, obstructive hypopnea, and obstructive sleep apnea can occur that eventuate in sudden death of children.[16]

Pediatric dentists were found to have a better outlook compared to oral maxillofacial surgeons with a response ratio of 2:1. This difference could be due to the emerging role of pediatric dentist in recent era in the therapeutic approach toward obstructive sleep apnea as a "sentinel" who can observe and detect early signs of the disease for immediate referral and also takes active participation in therapy.[17] Only 15.6% of specialists presume that there can be a chance of unexpected and unexplained asphyxia in

infants due to tethered oral tissues of which pediatric dentists have better standpoint in this context ($p = 0.002$).

During the swallowing mechanism, the posterior part and mid part of the tongue aid to prevent inhaling regurgitated stomach contents. In case of ankyloglossia, the infant must actively work to push the tongue back to modulate the opening between the airway and gastrointestinal tract. Once the infant falls asleep, the infant no longer actively works to protect the airway, thus allows regurgitated stomach contents to be inhaled leading to asphyxia. If an infant is having difficulty breastfeeding and a tongue tie is not obvious, evaluating the upper-lip frenum can help alert the clinician to the existence of a possible submucosal tongue tie. Haller and Brown postulated that ankyloglossia (tongue tie) and restricted upper lip (lip tie) might be maturational biologic vulnerabilities that contribute to unexpected and unexplained asphyxia with strong correlation.[18]

Majority of specialists (77.6%) conjectured that tongue ties in infancy could cause articulation issues and result in delayed speech with varied opinion between the specialties ($p < 0.05$). The tongue is very important in the formation of certain sounds and "phonemes" in speech. Ankyloglossia impairs the tongue mobility, which results in altered phonation.[19] Messner and Lalakea have documented articulation problems in 21 children with ankyloglossia by measuring tongue mobility, speech evaluation, and parental questionnaires. He showed improvement in tongue mobility and articulation in 71.4% children after revision of tongue tie. But, in case a patient articulates all those sounds properly, the surgical approach is not recommended.[20]

About 67.7% specialists believed that persistence of tethered oral tissues might lead to atypical swallowing with significant opinion difference between pediatric dentists and oral surgeons ($p = 0.187$). Natural breastfeeding plays an important role in the maturation of the perioral musculature, thus supports the development of a normal mature swallowing.[21] Ankyloglossia creates an imbalance between the perioral muscles as the tongue fails to elevate in order to create a seal and thus continues to thrust forward, subsequently resulting in atypical swallowing.[21,22]

Almost 71.4% specialists predicted that presence of tethered oral tissues can result in spacing between the teeth but only 19.3% specialists believed tethered tissues can cause gingival retraction. No significant difference exists between the opinions of pediatric dentists and oral surgeons ($p > 0.05$). Upper lip tie is a benign condition that tends to improve with normal facial growth, but a hyperplastic maxillary frena is associated with a diastema of the upper central incisors and traction of the attached gingiva.[23,24] The insertion of the lingual frenulum in the area of the papilla had the highest association with pathological retraction of the lingual marginal gingival tissue, thus creating periodontal defect with gingival recession and begins with the eruption of permanent incisor at the age of 6 years. If neglected, both soft tissue recession and bony defects have potential to compromise tooth retention.[25]

In the present study, 13.5% specialists believed that tied tissues can increase the caries susceptibility. This phenomenon was reported when the lip attachment ingress into the zone where the upper centrals will emerge, classified as class III lip tie, and also in situations where the close attachment extends beyond the maxillary alveolar ridge into the palatal area, classified as a class IV lip tie.[26] This close approximation causes ineffective latching or suckling, hence may take longer feeding time, increased frequency of feeds, often provide a pocket sulcus for residual milk and food debris, resulting in increased caries risk.[7,26] Also, severely tethered tongue limits the normal cleansing action, subsequently resulting in increased exposure to residual milk.[7] Kotlow has observed a peculiar caries pattern with significant notching on the incisal facial surface in class III or class IV lip tie infants.[26]

In the present study, almost 43.2% specialists with opinions of both pediatric dentists and oral surgeons in 1:1 ratio shrewd enough to anticipate the occurrence of malocclusion in the infants with severe tongue tie/lip tie. During jaws development, the tongue acts as scaffold stimulating a normal growth of the upper jaw. But in case of ankyloglossia, the tongue has to descend and protrude. So, the pressure of unbalanced perioral structures constricts the maxillary arch, resulting in narrow, V-shaped and high vault palate with maxillary hypodevelopment.[3] Defabianis stated that ankyloglossia prevents proper lip seal during swallowing, leading to tongue thrusting, which in turn can cause open bite. Reduction of mandibular intercanine width can occur due to pull of the short lingual frenum resulting in constriction of the mandibular anterior region.[27] Mandibular prognathism can also develop as a sequel of exaggerated tongue thrusting.[3,28] Thus, tethered oral tissues significantly affect the development of the maxilla and mandible resulting in malocclusion. Only 10.4% specialists of the target population believed that untreated tethered tissues can result in postural alteration in future. The persistence of morphofunctional alterations due to tethered oral tissues can strain suprahyoid musculature, subhyoid musculature, and spinal column, resulting in anteriorization of the head and posterior protrusion of the shoulder.[29]

While the present study has provided some important information on sequelae of tethered oral tissues in infancy, it has some limitations. The present study was unable to determine attitude of specialty dentists toward management of tethered oral tissues. Further studies should include these aspects into consideration and explore more factors affecting the utilization of dental services in this population in depth. The other limitation observed was a small sample size, hence further investigations should include a large number of subjects to make the results more generalizable.

Conclusion

Most specialists in this study are found knowledgeable to predict functional limitations caused by tethered oral tissues such as breastfeeding issues (71.9%), articulation defects (77.6%), and development of atypical swallowing (67.7%). But limited awareness exists regarding the other sequelae of tethered orofacial structures such as deviant breathing patterns causing sleep disturbances, pathological retraction of gingival tissues and increased susceptibility to caries, morphological dentoskeletal alterations, and postural defects.

Infant oral health care professionals like pediatric dentists, being the frontline oral health care providers for children from infancy through adolescence, should understand the anatomical constrains caused due to tethered tissues in infancy that can have dramatic impact on human physiology and quality of life. Diagnosis and management of tethered oral tissues in infancy requires related comprehensive theoretical knowledge and diverse collection of skills, respectively. Hence, there exists a need to reform the curriculum with an elaborative view on tethered oral attachments and their sequelae in accordance with current disease (tongue tie/

lip tie) demographics and changing treatment demands that form the basis of critical thinking, lifelong learning, clinical reasoning, and judgment.

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