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Association of Short Lingual Frenum with Obstructive Sleep Apnea: A Systematic Review and Meta Analysis

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Abstract

Introduction: Recent evidences have emphasized the role of a short lingual frenulum in the pathogenesis of sleep disordered breathing. The oral dysfunction induced by a short frenulum may promote oral-facial dysmorphism decreasing the size of upper airway lumen and increasing the of upper airway collapsibility during sleep.

Aim: The aim of this systematic review was to assess whether there is sufficient evidence to suggest that short lingual frenum contribute to the development of obstructive sleep apnea.

Methodology: This review included observational and interventional studies that assessed the lingual frenulum in children with sleep disordered breathing were included. Electronic data bases included PubMed, MEDLINE, EBSCO, Google Scholar etc. Search strategy consisted of words like Ankyloglossia; sleep apnea; short lingual frenum etc. Following the selection of studies, pertinent data was extracted and subjected to qualitative and quantitative synthesis. The risk of bias was appraised based on established methodologies and heterogeneity was assessed.

Conclusion: According to this systematic review an association of short lingual frenum with obstructive sleep apnea can be observed in most of the cases. Children with obstructive sleep apnea should be evaluated for a short lingual frenulum, and vice versa.

Keywords: Short Lingual Frenum, Intermittent, Ankyloglossia, Sleep Apnea.

Introduction

Obstructive sleep apnea syndrome is defined by the American Thoracic Society (ATS) as "A disorder of breathing during sleep characterized by prolonged partial upper airway obstruction and/or intermittent complete obstruction (obstructive apnea) that disrupts normal ventilation during sleep and normal sleep patterns". The prevalence in children is estimated in the range of 2%–

4% from western countries. According to studies the prevalence of Paediatric OSAS in India is about 9.6%. The age group in which it is commonly seen is between 2-6 years. It has shown adverse effects on childhood behaviour, neurodevelopment, metabolism, and overall health. Various risk factors of OSA in children include obesity, adenoid and tonsil hypertrophy, allergic rhinitis and septal deviation. Some studies have shown that a short lingual frenulum may also contribute to the development of OSAS in children.[1]

Short lingual frenum (Ankyloglossia) is an anatomical condition characterized by the restriction of tongue movement, affecting its function, the shape of the dental arches, and their consequent occlusion. It is a congenital anomaly seen in 4-5% of the population. Incomplete migration or no migration of cells of lingual frenum results into short lingual frenum. Recent studies have shown that the short lingual frenum may contribute to development of OSAS in children. In a report, Guilleminault et al concluded that if short lingual frenum is kept untreated is associated with OSAS at a later stage.[3]

The literature on the correlation between ankyloglossia and OSA is scarce, as shown in a systematic review conducted by Chinnadurai et al. in 2015.[2]

Aim: The aim of this systematic review was to assess the association of short lingual frenum with obstructive sleep apnea in children.

Materials and Methods

Inclusion and exclusion criteria

This systematic review has included studies in which children are associated with short lingual frenum and also the studies in which children are diagnosed with obstructive sleep apnea. Studies providing information about association between short lingual frenum and obstructive sleep apnoea in terms of odds ratio, number of events occurring in two groups were also included. The studies included where published in the English language only till the year March 2023.

The studies included were cross sectional studies, RCTs and retrospective studies.

The exclusion criteria consisted of studies involving diseases other than obstructive sleep apnoea. Review reports, case series, in-vitro and animal studies were also excluded.

Search strategy: A systematic review of literature and meta-analysis was performed. This study followed the (PRISMA 2020) Preferred Reporting Items for Systematic Review 2020[17], the Cochrane Handbook for systematic reviews of interventions, version 5.1.0. and 4th Edition of the JBI Reviewer's Manual and was registered at PROSPERO under registration code CRD42023396359**.**

Primary outcomes measured were events of occurrence of obstructive sleep apnoea among participants with short lingual frenum. The preferred reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) for conducting a meta-analysis were followed. The electronic data resources consulted for elaborate search were PubMed, Google scholar, Directory of Open Access Journals (DOAJ).

Studies were selected based on the PEOS inclusion criteria in the review protocol. Two reviewers assessed titles and abstracts to identify potentially eligible studies. Any queries were discussed with a third reviewer. The search provided a total of 2040 articles. After removing the duplicates and screening the total articles obtained were 256. The full text articles after assessing for the eligibility were found to be 45. Studies which were included in qualitative synthesis were 4. And the studies included in the quantitative synthesis were 3.

Literature Search and Search Strategy

Study selection: Two reviewers independently screened all the retrieved titles and abstracts. If one reviewer considered one publication as having met the inclusion criteria, the full text was obtained. Abstracts considered as potentially eligible, as well as those with no enough information, were considered for full text assessment. Any differences regarding eligibility were resolved through consensus.

Data extraction: Two reviewers independently extracted data from the included studies. Disagreements were again resolved through discussion. Data gathered was carried out using a verification list of items that were considered for data extraction. Details regarding the publication and the study, the participants, settings, the interventions, the comparators, the outcome measures, study design, statistical analysis and results, and all other relevant data (funding; conflict of interest etc.) were carefully and accurately extracted from all included studies. Data extraction was done and accurately recorded in the excel sheets for all the primary outcomes separately. Table 2 shows reviewed treatment outcome.

Risk of bias: Table 3 shows risk of bias for this systematic review. Quality Assessment of the selected studies was performed using Newcastle Ottawa tool for quality assessment of cross-sectional studies and retrospective studies.[18]This scale includes 3 main categories: selection of study groups, comparability, and outcome.

Each individual study received a maximum of 9 points. In case of no consensus during the evaluation process, discrepancies were discussed with a third reviewer.

Results

The initial electronic database search on PubMed/MEDLINE, Cochrane library and DOAJ resulted in 2065 titles. Two hundred ninety-eight articles were cited as duplicates. After screening the abstracts, 256 relevant titles were selected by two independent reviewers and 1511 were excluded for not being related to the topic. Following examination and discussion by the reviewers, 45 articles were selected for full-text evaluation. Hand searching of the reference lists of the selected studies did not deliver additional papers. After pre-screening, application of the inclusion and exclusion criteria and handling of the PEOS questions, five studies remained. Five studies were included in the qualitative synthesis, three out of which were subjected for data extraction and statistical analysis. **(Figure 1)**

Study Characteristics:

These studies were conducted in different parts of world, with two studies[3,4] in USA, one in Italy[4], one in Poland[13] and one[21] in Hongkong. Two studies[4,13] provided information about ethical approval for the study. The study design for 2 studies was retrospective[3,7], three were cross-sectional[4,13,21]. In most of the studies, evaluation of obstructive sleep apnea was done using Pediatric sleep questionnaire (PSQ), Friedman scale, Mallampati scale, facial harmony, sleep clinical record (SCR), etc.

Evaluation of short lingual frenum was done using free tongue length test. Overall, 895 children within age group of 2-17 years participated in the study.

The conclusions of all the studies revealed that short lingual frenum is a risk factor of sleep apnea. (Table 2)

Risk of Bias

Table 3 and 4 depicts risk of bias according to Newcastle Ottawa tool for retrospective studies and cross-sectional studies.[19]

Among the two retrospective studies, both studies showed low risk of bias[3,7]. In case of cross-sectional

studies, two showed moderate risk[4,13] and one[21] showed low risk of bias.

Meta-analysis

Meta-analysis was conducted on three studies based on following parameters:

1. Children with obstructive sleep apnea having short lingual frenum
2. Length of free tongue in children with and without sleep apnoea

Odds ratio for association between short lingual frenum and obstructive sleep apnea

Two studies[3,4] were included to obtain pooled odds ratio. The pooled odds ratio was 3.40 [1.94, 5.95] indicating that the odds of occurrence of obstructive sleep apnea were greater with short lingual frenum as compared to control group.

As heterogeneity (I^2) was 0%, fixed effects model was used. Overall these results were statistically significant. Refer figure number 2.

Length of free tongue

Two studies[3,13] provided data regarding length of free tongue in case and control groups. Pooled mean difference was -2.20 [-3.47, -0.93] which indicated that the length of free tongue was less in cases of obstructive sleep apnea as compared to controls. Refer figure number 3.

Discussion

Systematic investigation of a short frenulum at birth and clipping in early infancy varies greatly depending on the geographic location of delivery.[3] The development of the maxilla is notably impacted by a short lingual frenulum because of the tongue's low position. In addition to mouth breathing while you sleep, it causes the abnormal growth of a high, narrow hard palate. Since the first two years of life are when the orofacial growth is particularly rapid, these changes happen early in life.[3,7] Recognition of a short lingual frenulum in a toddler or older kid should prompt assessment of SDB because it is known that subsequent anatomical alterations in the oral cavity enhance the degree of collapsibility of the upper airway during sleep.[7]

The present systematic review was done to evaluate the association of short lingual frenum with obstructive sleep apnea in children. The review identified four studies assessing the association between short lingual frenum and OSA. There is an agreement between the four analyzed studies that the lingual frenulum alteration interferes with the growth of anatomical structures, leading to alterations in respiratory, suction, chewing and speech functions.

The overall results from meta-analysis depicted the strong statistically significant association of short lingual frenum and obstructive sleep apnea. Additionally treating the short lingual frenum might reduce the occurrence of apnea. The studies by Zaghi et al[22] and Baxter et al.[10] showed that the association of myofunctional therapy with lingual frenectomy brought benefits to treated patients, showing the importance of a multidisciplinary action.

However, there's still a need for further prospective research studies to compare different diagnostic performances of these models and to help in the extrapolation of paediatric OSA. This is due to the lack of similarity in the signs and symptoms narrated by all these above-mentioned studies and the limited research in this field of study. The new prospective studies need to be designed in a way to highlight every specific symptom and indicator that this systematic review revealed, such as EDS, sleep apnoea, and respiratory difficulties when sleeping, as well as highly sensitive symptoms like wheezing and tonsillar size. This

combined approach could boost sensitivity while maintaining a reasonable level of specificity.

Conclusion

1. The findings analysed in this review support the hypothesis that obstructive sleep apnea and ankyloglossia are associated and short lingual frenum.
2. It can be considered as an important clinical sign in diagnosing obstructive sleep apnea.
3. According to the findings of these research, a tongue with limited mobility can influence speech, mastication, respiration, and sleep, and that its appropriate surgical release combined with myofunctional treatment can lead to an increase in function and quality of life.

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Legend Figures & Tables

Figure 1: PRISMA flow diagram

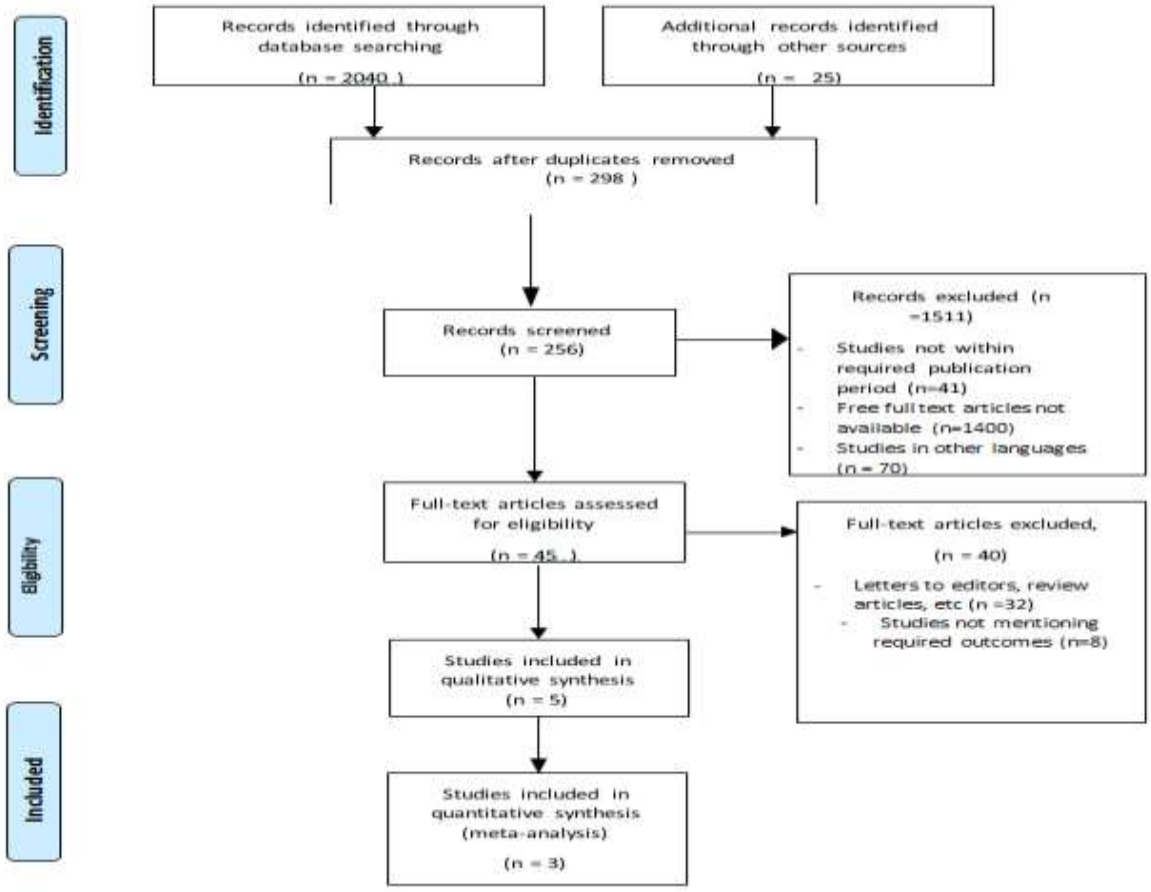

Figure 2:

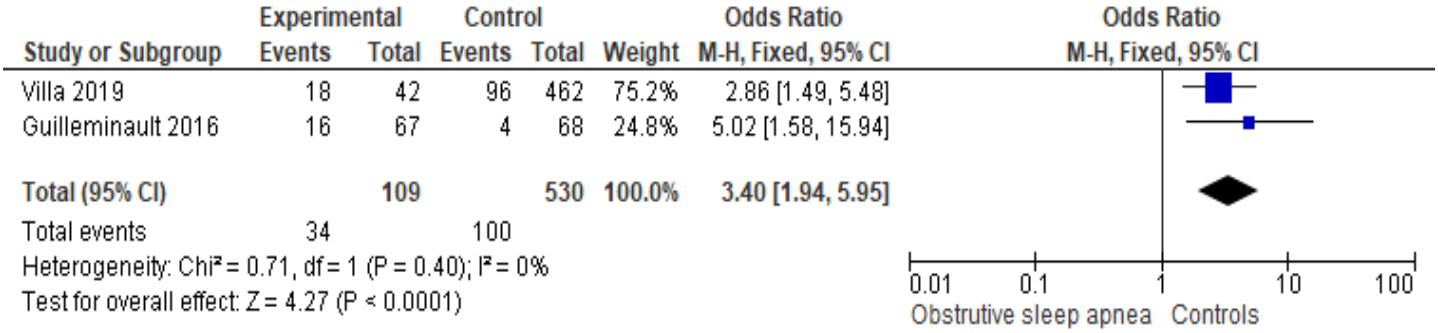

Figure 3:

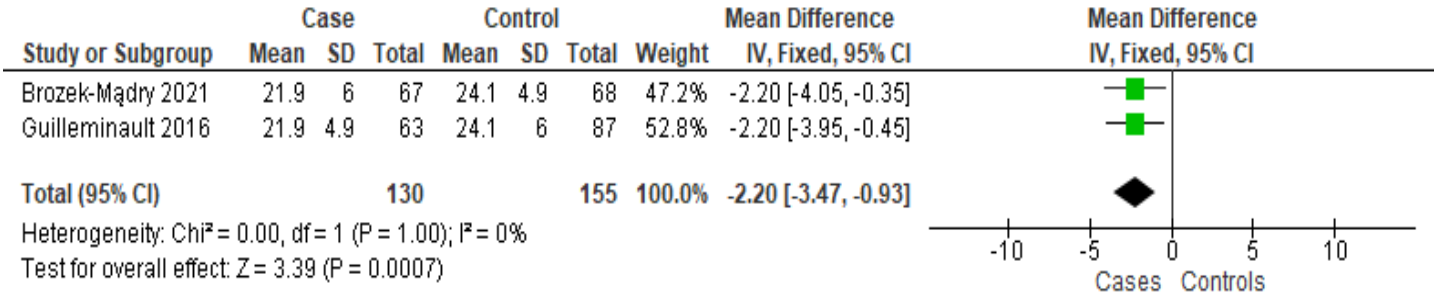

Table 1: Concept table

Population	("child"[MeSH Terms] OR "child"[All Fields] OR "children"[All Fields]) AND (short[All Fields] AND ("lingual fraenum"[All Fields] OR "lingual frenum"[MeSH Terms] OR ("lingual"[All Fields] AND "frenum"[All Fields]) OR "lingual frenum"[All Fields]))
Exposure	((("obstructive sleep apnoea"[All Fields] OR "sleep apnea, obstructive"[MeSH Terms] OR ("sleep"[All Fields] AND "apnea"[All Fields] AND "obstructive"[All Fields]) OR "obstructive sleep apnea"[All Fields] OR ("obstructive"[All Fields] AND "sleep"[All Fields] AND "apnea"[All Fields])) OR (obstructive[All Fields] AND ("sleep wake disorders"[MeSH Terms] OR ("sleep"[All Fields] AND "wake"[All Fields] AND "disorders"[All Fields]) OR "sleep wake disorders"[All Fields] OR ("sleep"[All Fields] AND "disorder"[All Fields]) OR "sleep disorder"[All Fields]))) OR ("sleep apnoea"[All Fields] OR "sleep apnea syndromes"[MeSH Terms] OR ("sleep"[All Fields] AND "apnea"[All Fields] AND "syndromes"[All Fields]) OR "sleep apnea syndromes"[All Fields] OR ("sleep"[All Fields] AND "apnea"[All Fields]) OR "sleep apnea"[All Fields])) AND ("child"[MeSH Terms] OR "child"[All Fields] OR "children"[All Fields])
Outcome	((obstructive[All Fields] AND ("sleep wake disorders"[MeSH Terms] OR ("sleep"[All Fields] AND "wake"[All Fields] AND "disorders"[All Fields]) OR "sleep wake disorders"[All Fields] OR ("sleep"[All Fields] AND "disorder"[All Fields]) OR "sleep disorder"[All Fields])) AND (short[All Fields] AND ("lingual fraenum"[All Fields] OR "lingual frenum"[MeSH Terms] OR ("lingual"[All Fields] AND "frenum"[All Fields]) OR "lingual frenum"[All Fields]))) AND ("association"[MeSH Terms] OR "association"[All Fields])
Study design	((crossectional[All Fields] AND "studies"[All Fields]) AND ("randomized controlled trial"[All Fields] OR "randomized controlled trials as topic"[MeSH Terms] OR "randomized controlled trials"[All Fields] OR "randomised controlled trials"[All Fields])) AND ("retrospective studies"[MeSH Terms] OR ("retrospective"[All Fields] AND "studies"[All Fields]) OR "retrospective studies"[All Fields] OR ("retrospective"[All Fields] AND "study"[All Fields]) OR "retrospective study"[All Fields])

Table 2: Characteristics of included studies

Sr no	Study Id	Study Design	Country	Ethical Approval	Sample Size		Age	Gender	Evaluation Of Sleep Apnea	Evaluation Of Frenum	Conclusion
					Study Group	Control Group					
1	Huang 2015	retrospective study	USA	not mentioned	27		2-17	M 18, F 9	PSQ, Friedman scale, Mallampati scale, facial harmony	-	Children with SDB should be evaluated for a short lingual frenulum, and conversely, children with an abnormally short frenulum should be investigated for the presence of SDB
2	Guilleminault 2016	retrospective study	USA	not mentioned	63	87	3-12	M 92, F 58	PSQ, pediatric day time sleepiness scale, Friedman scale, Mallampati scale, facial harmony	free tongue length	A short lingual frenulum left untreated at birth is associated with OSAS at later age, and a systematic screening for the syndrome should be conducted when this anatomical abnormality is recognised.
3	Villa 2019	Cross-sectional	Italy	yes	504		6-12	M 199, F 23	Sleep clinical record	free tongue length, tongue pressure	Short lingual frenulum is a risk factor for SDB. An early multidisciplinary approach and screening for SDB are indicated when this anatomical abnormality is recognized.
4	Brozek-Mądry 2021	Cross-sectional		yes	67	68	4-17	M 65, F 70	PSQ, head forward posture	Quick tongue tie assessment tool, length of free tongue	The study identified a relationship between a short lingual frenulum and the risk of OSAS in children. Detecting and addressing ankyloglossia in children is necessary before it leads to orofacial changes, malocclusion, and consequently, sleep apnea
5	Yuen 2021	Cross-sectional	Hongkong	not mentioned	50	36	5-12	-	obstructive apnea hypopnea index (OAHI)	tongue mobility, free tongue length	Reduced tongue mobility is associated with OSA in prepubertal children. Furthermore, tongue mobility may be an important factor in driving mandibular development.

Table 3: Newcastle Ottawa scale for risk of bias of case-control studies

Selection				Comparability		Outcome			Total score	Risk
Case definition	Representativeness of case	Selection of controls	Definition of controls	Main factor	Additional factor	Ascertainment of exposure	Same method of ascertainment for cases and controls	Non-response rate		
*	*	*	-	*	-	*	*	*	7	Low risk
*	*	*	*	*	-	*	*	*	7	Low risk

Table 4: Newcastle Ottawa scale for risk of bias of Cross-sectional studies

Selection				**Comparability**		**Outcome**		**Total score**
Representativeness of sample	Sample size	Non-responders	Ascertainment of exposure	Main factor	Additional factor	Assessment of outcome	Statistical test	
*	-	*	*	-	-	*	*	5
*	-	-	*	*	-	*	*	5
*	-	*	*	*	-	*	*	6