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## Background

- **Ankyloglossia (tongue-tie)** is a clinical diagnosis characterized by a congenitally short lingual frenulum thought to restrict tongue movement.
- The relationship between the structure of the lingual frenulum and functional limitations in children remains uncertain.
- The **uncertainty** surrounding the **association between ankyloglossia and pediatric sleep disordered breathing (SDB)**, including obstructive sleep apnea (OSA), has led to confusion and management controversies among clinicians and parents.
- Currently, invasive procedures to release the lingual frenulum, including frenectomy and frenotomy, are offered by some clinicians for the treatment of SDB in children with a diagnosis of ankyloglossia. However, **the use of invasive procedures with the goal of improving SDB warrants further research.**
- With the **increasing prevalence of ankyloglossia diagnosis** and frenectomies performed, a better understanding of the relationship between ankyloglossia and SDB is imperative.

## Objectives

1. To examine the relationship between ankyloglossia and SDB
2. To assess the impact of frenulotomy/frenulectomy on the disease course of SDB in pediatric patients.

## Methods

- This systematic review was conducted in accordance with the PRISMA guidelines.
- **2 independent reviewers** screened studies for inclusion, extracted data, and performed risk of bias assessment.

### Screening

- English literature assessing the relationship between ankyloglossia and SDB in non-syndromic children
- **Inception to August 16, 2022:** EMBASE, Web of Science, Medline, CINAHL, CCRCT and SCOPUS

### Inclusion

Case series, retrospective cohort studies, case control studies, prospective cohort studies, randomized control trials

### Exclusion

Opinion papers, technical reports, editorials, literature reviews and case reports

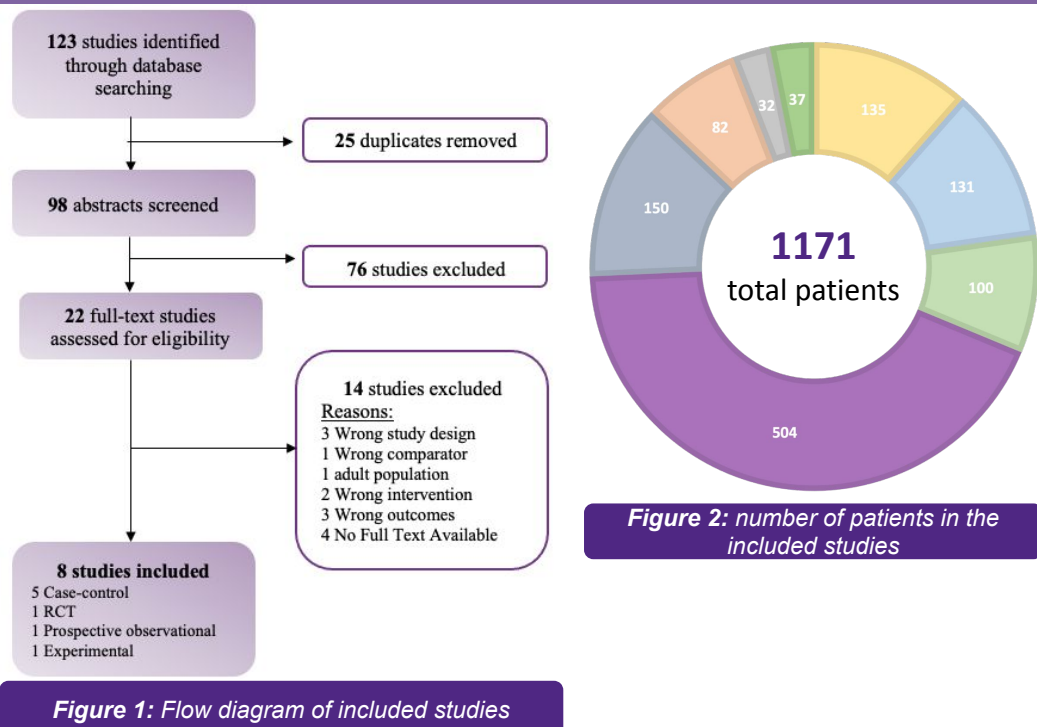
- Pediatric patients (<18 years)
- Diagnosis of ankyloglossia
- Patients are being assessed for SDB
- Patients with or without surgical treatment (frenulotomy, frenulectomy, frenuloplasty), medical treatment, speech therapy and osteopathy

Syndromic patients (i.e. those with Trisomy 21, craniofacial abnormalities, Pierre Robin Sequence, etc.)

### Data Extraction & Analysis

- A standardized data collection sheet was developed a priori
- Risk of Bias Assessment: Cochrane risk of bias, Newcastle risk of bias

## Results



Author, year	Country	Design	Inclusion Criteria	Measure of ankyloglossia	Measure of SDB	Patients, n	Definition	Association between ankyloglossia & SDB
Brożek-Mądry, 2021	Poland	Case-control	Children living in the rural areas of Poland	Length of free tongue ≤16mm	PSQ	67	Risk of OSAS (PSQ≥8)	+
						68	No risk of OSAS (PSQ<8)	
Burska, 2022	Poland	Case-Control	Non-syndromic children living in rural areas of central Poland.	Length of free tongue ≤16mm	PSQ	65	Risk of SDB (PSQ≥8)	+
						66	No risk of SDB (PSQ<8)	
Cohen-Levy, 2020	Canada	Prospective Observational	Non-syndromic symptomatic children who had previously undergone surgery of adenoids and/or tonsils reporting chronic snoring or witnessed apneas	Limited tongue elevation in relation to maximal aperture of 60% or less	Self-reported questionnaire (HSCS); Sleep clinical record (SCR); Home sleep apnea test (HSAT)	100	chronic snoring (HSCS > 0) or suspected OSA (HSCS > 2.72) undergoing SCR test and/or HSAT	No association
Villa 2019	Italy, Greece	Case-Control	Non-syndromic children recruited from a school in Rome who correctly filled out the SCR questionnaire	Length of free tongue ≤16mm	Sleep Clinical Record (SCR)	42	SCR ≥ 6.5	+
						462	SCR < 6.5	
Guilleminault, 2016	USA	Case-Control	Non syndromic children referred to a clinic for "sleep disorders".	Length of free tongue ≤16mm	PSQ; paediatric daytime sleepiness scale. PSG to confirm.	63	Short lingual frenulum	+
						87	Normal frenulum	
Yuen, 2021	Hong Kong	Case-Control	Prepubertal children suspected to have OSA	Length of free tongue ≤16mm	PSG; Sleep questionnaires (OSA-18, Hong Kong Children sleep questionnaire (HK-CSQ))	48	positive OSA (DAHI ≥ 1/hr)	No association
						34	no OSA	
Fioravanti, 2021	Italy	RCT	A diagnosis of OSAS and a short lingual frenulum, the presence of a pathological lingual frenulum (class III-IV-V by Kotlow; Grades 2 and 3 by Ruffoli)	Length of free tongue ≤16mm	PSG	16	Frenectomy and myofunctional + speech therapy	+
						16	Myofunctional + speech therapy only	
Baxter 2020	USA	Experimental	Children with speech, feeding, and sleep issues, diagnosis of symptomatic tongue restriction	Unclear	Non-standardized parental reports	37	Underwent frenectomy	+

Table 1: Showcases characteristics and findings of all included studies.

## Risk of Bias Assessment

Cochrane Risk of Bias: Randomized Control Trial							
Q1	Q2	Q3	Q4	Q5	Q6	Q7	
Fioravanti 2021	+	+	?	-	-	-	?

Q1: Selection Bias: Random Sequence Allocation  
 Q2: Selection Bias: Allocation Concealment  
 Q3: Reporting bias: Selective reporting  
 Q4: Performance Bias: blinding of participants and personnel  
 Q5: Detection Bias: blinding of outcome assessment  
 Q6: Attrition Bias: Incomplete outcome data  
 Q7: Other Bias

Newcastle Risk of Bias: Case-Control Studies									
	S1	S2	S3	S4	C1	E1	E2	E3	Quality
Brożek-Mądry, 2021	0	★	★	0	★	0	★	-	fair
Burska, 2022	0	★	★	0	★	0	★	-	fair
Guilleminault, 2016	0	0	★	0	★	0	★	-	poor
Yuen, 2022	0	★	0	★	★	0	★	-	fair
Villa, 2019	0	0	★	0	★	0	★	-	poor
Maximum allotment	1	1	1	1	2	1	1	1	Good

S1: Is the case definition adequate?  
 S2: Representativeness of the cases  
 S3: Selection of Controls  
 S4: Definition of Controls  
 C1: Comparability of cases and controls on the basis of the design or analysis  
 E1: Ascertainment of exposure  
 E2: Same method of ascertainment for cases and controls  
 E3: Non-Response rate

Table 2: Risk of bias assessment using Cochrane risk of bias for RCTs and Newcastle risk of bias tool for Case-Control Studies included in our study

## Summary of Results

- 8 studies (5 case-control, 1 randomized control trial (RCT), 1 experimental and 1 prospective observational), involving 1171 patients in total, were included
- Sample size across studies range from 32 to 504, with age ranging from 13 months to 17 years.
- **Conflict of Interest:** 4 studies declared no financial conflicts or competing interests, 3 studies reported funding, and 1 study did not report such disclosures.
- **SDB outcome measurements:** 3 studies used the gold standard (PSG), 1 study used HSAT (similar accuracy to PSG), 3 studies used PSQ and 2 studies used SCR. 1 study used non-standardized parental reports, and 1 study used HK-CSQ and OSA-18 questionnaires.

## Conclusion

- A **definitive conclusion** on the **association between ankyloglossia and SDB cannot be made due** to significant risk of bias and high heterogeneity across all studies.
- **Frenotomy should not routinely** be recommended to improve SDB in children without further research.
- Further research to better understand the relationship between ankyloglossia and SDB is imperative for informing clinicians and implementing practice guidelines when children with a diagnosis of ankyloglossia present with SDB.

## Discussion

- 2 studies reported improvement of SDB symptoms following frenectomy.
- 2 studies did not report an association between a short lingual frenulum and SDB, whereas 3 studies did. 1 study reported an association with univariate analysis only.
- A risk of bias assessment was performed on 5 cross-sectional case-controls and 1 RCT; there is a significant concern for bias across these studies.
- A risk of bias assessment could not be performed on 3 studies (Baxter et al, Cohen-Levy et al, and Fioravanti et al) as they did not follow case-control, cohort or RCTs study designs.
- However, there are concerns of unclear ankyloglossia and SDB outcome methods and measurements, unblinded assessors, and non-randomized patient selection across all studies

### LIMITATIONS OF STUDIES INCLUDED:

- (1) Non-standardized measures and diagnosis of ankyloglossia across studies
- (2) Most studies did not utilize the gold standard measures of OSA (PSG)
- (3) High heterogeneity within and between groups with non-standardized reporting of variables and outcomes

## References

1. Mills N, Keough N, Geddes DT, Pransky SM, Mirjalili SA. Defining the anatomy of the neonatal lingual frenulum. *Clinical Anatomy*. 2019 Sep;32(6):824-35
2. Messner AH, Walsh J, Rosenfeld RM, Schwartz SR, Ishman SL, Baldassari C, Brietzke SE, Darrow DH, Goldstein N, Levi J, Meyer AK. Clinical consensus statement: ankyloglossia in children. *Otolaryngology-Head and Neck Surgery*. 2020 May;162(5):597-611
3. Walsh J, Links A, Boss E, Tunkel D. Ankyloglossia and lingual frenotomy: national trends in inpatient diagnosis and management in the United States, 1997-2012. *Otolaryngol Head Neck Surg*. 2017;156(4):735-740.
4. Segal LM, Stephenson R, Dawes M, Feldman P. Prevalence, diagnosis, and treatment of ankyloglossia: methodologic review. *Canadian Family Physician*. 2007 Jun 1;53(6):1027-33.
5. Lisonek M, Liu S, Dzakupasu S, Moore AM, Joseph KS, Canadian Perinatal Surveillance System (Public Health Agency of Canada). Changes in the incidence and surgical treatment of ankyloglossia in Canada. *Paediatrics & child health*. 2017 Oct 1;22(7):382-6