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# Short lingual frenulum as a risk factor for sleep-disordered breathing in school-age children

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**ABSTRACT**

**Background:** Recent evidence has emphasized the role of a short lingual frenulum in the pathogenesis of sleep-disordered breathing (SDB) in childhood. The oral dysfunction induced by a short frenulum may promote oral−facial dysmorphism, decreasing the size of upper airway lumen and increasing the risk of upper airway collapsibility during sleep. The aim of this study was to evaluate the presence of a short lingual frenulum as risk factor for SDB in children of school age, with and without snoring, who were recruited from the community.

**Methods:** Children aged 6−14 years were recruited from a school in Rome. For all participants, the previously described Sleep Clinical Record (SCR) was completed, and orthodontic evaluation and measurement of lingual frenulum were performed. Tongue strength and endurance were evaluated in all participants using the Iowa Oral Performance Instrument (IOPI). SDB was defined as positive SCR (≥6.5).

**Results:** We assessed 504 children with mean age of 9.6 ± 2.3 years, and in 114 of them (22.6%) a short frenulum was identified. Children with a short lingual frenulum were at significantly higher risk for a positive SCR compared to those with a frenulum of normal length (odds ratio = 2.980, 95% confidence interval = 1.260−6.997). Participants with positive or negative SCR did not differ in tongue strength or endurance.

**Conclusion:** 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.

## 1. Introduction

Obstructive sleep-disordered breathing (SDB) is associated with increased upper airway resistance and abnormal pharyngeal collapsibility, leading to partial or complete upper airway obstruction during sleep [1,2]. Fat tissue deposition in the upper airway wall, nonfat tissue−related hypertrophy of the upper airway structures, and special craniofacial features, in combination with genetic and environmental factors, may contribute to restriction of the retropalatal and retroglossal space [3−5].

Several authors have described associations between malocclusion and functional disorders of the oral cavity and the adjacent muscles [6]. Abnormal mandibular development and malocclusion can affect the respiratory function by reducing the upper airway size [7,8]. Children with habitual snoring and SDB frequently experience delayed mandibular growth, leading to mandibular retroposition and posterior displacement of the tongue [9−11]. Orthodontic treatment can improve dental as well as maxillary and mandibular alignments, facilitating enlargement of the upper airway and resolution of SDB [8,12].

Ankyloglossia, or shortening of the free lingual portion, is an anatomical condition characterized by the restriction of tongue movement, affecting its function, the shape of the dental arches, and their consequent occlusion [13]. During tongue embryogenesis, the cells of the lingual frenulum undergo apoptosis and migrate distally to the medial region of the lingual dorsum. Ankyloglossia is due to incomplete migration or no migration of the lingual frenulum cellsm which results in a short lingual frenulum. In a recent report, Guilleminault et al investigated retrospectively 150 successive children with obstructive sleep apnea syndrome (OSAS), 63 of whom were shown to have a short frenulum, concluding that a short frenulum left untreated at birth is associated with OSAS at a later age [1]. A second paper from the same group reported on 150 successive children studied at the time of OSAS investigation, and indicated that 42% of them had short frenulum that was always associated with abnormal development of the oral cavity early in life [14]. Thus, the aim of this study was to evaluate the presence of a short lingual frenulum as a risk factor for SDB in children of school age, with and without snoring, who were recruited from the community.

## 2. Patients and Methods

### *2.1. Participants*

The Sapienza University Institutional Review Board approved the study, and written informed consent was obtained from the parents of children upon recruitment. Children aged 6−14 years were prospectively recruited from a school in Rome. Exclusion criteria were as follows: a positive history of acute or chronic cardiorespiratory or neuromuscular disease; chronic inflammatory disease; major craniofacial abnormalities; chromosomal syndromes; and epilepsy.



### *2.2. Evaluation for SDB*

For each participant, a medical history was obtained and a physical examination was performed for symptoms and signs related to SDB, and the Sleep Clinical Record (SCR) was completed. Details of the SCR, a clinical tool for the detection of children at high risk for SDB, have been described elsewhere [15]. In brief, a total SCR score is calculated after considering the following: abnormalities in the nose, oropharynx, dental, and craniofacial occlusion; Brouillette OSAS score [16]; and presence of symptoms of inattention and hyperactivity. A total SCR score ≥6.5 is considered positive, and it is associated with high risk of OSAS defined as an obstructive apnea−hypopnea index (AHI) >1 episode/h) [15].

More specifically, the dental/skeletal malocclusion included jaw deviation from normal occlusion, such as retrognathia, prognathia, open bite, deep bite, crossbite, overbite, and overjet. An overjet value >4.0 mm was considered to be increased, and the normal overbite was determined to be one-third coverage of the lower incisors by the upper incisors. The term open bite was used in cases without vertical overlap or contact between the anterior incisors when the jaw was closed. Crossbite described the buccal version of the mandibular teeth relative to the maxillary teeth. High-arched palate was defined as an abnormally pronounced curvature angled superiorly along the palatal midline.

Severity of nasal septum deviation was classified in 3 categories. In grade I, the deviated septum did not reach the lower nasal turbinate; in grade II, the deviated septum reached the lower nasal turbinate; and in grade III, the deviated septum reached the lateral wall and compressed the lower nasal turbinate. In the SCR, the presence of the above abnormalities was scored with either 2 points (positive sign) or 0 points (negative sign).

*2.3. Evaluation of tongue strength and endurance*

To characterize the frenulum of a participant as being of normal length, we measured the free tongue length [17]. Kotlow defined the free tongue length as the distance between the insertion of the lingual frenulum into the base of the tongue and the tip of the tongue. The frenulum is considered short if the free tongue length is ≤16 mm [17].

Tongue characteristics may also affect the risk of OSAS, and they were evaluated using the Iowa Oral Performance Instrument (IOPI; IOPI Medical) [18]. The IOPI objectively measures tongue strength and endurance [18]. Tongue strength is expressed as the maximum pressure (in kPa) exerted when an individual puts pressure with her tongue on a disposable, standard-sized tongue bulb against the roof of the mouth. The standard procedure and reference values for age have been previously validated by Potter et al [19]. Maximum tongue pressure was measured in triplicate, with each effort lasting approximately 1 second and with a rest period of 1 minute between trials. Endurance was measured by asking subjects to maintain 50% of their maximum pressure for as long as possible, and the duration of the endurance measurement was expressed in seconds.

*2.4. Statistical analysis*

Positive SCR was the primary outcome measure, and short lingual frenulum was the main explanatory variable. Children without and with positive SCR were compared regarding the lingual frenulum length, frequency of a short lingual frenulum, and other variables that may affect the risk of OSAS and that are not included in the SCR, namely age, sex, body mass index z-score, obesity, tongue strength, tongue endurance. To evaluate the association between a short lingual frenulum and risk of OSAS, multivariable logistic regression was used to calculate the odds ratio and 95% confidence interval for having a positive SCR in children with versus without a short frenulum after adjustment for age, sex, tongue strength, and presence of obesity.

All variables were tested for normality. Accordingly, variables are expressed as a number and percentage (%) for categorical variables, mean ± standard deviation (SD) for normally distributed continuous variables, or median and interquartile range (25th−75th percentile) for non−normally distributed continuous variables. A $\chi^2$ test or Fisher exact test was applied for the comparisons involving categorical variables, and the independent *t* test or Mann−Whitney test for comparisons regarding continuous variables according to the normal distribution of the data. All the tests were 2-tailed, and a *P* value <0.05 was considered significant. The SPSS package PASW Statistics for Windows, Version 23.0) (SPSS Inc, Chicago, IL) was used for all of the analyses.

## 3. Results

### *3.1. Participant characteristics*

A total of 532 children were recruited initially; of these, 28 children were not evaluated because they were fulfilled one or more of the exclusion criteria. Thus, 504 children with a mean age of 9.6 (± 2.3) years were evaluated, 11.7% of whom were obese. Of 504 children, 42 (8.3%) were at high risk for OSAS as indicated by an SCR score ≥6.5. Abnormalities possibly contributing to upper airway obstruction in children without and with positive SCR are presented in Tables 1 and 2.

Children with positive SCR had higher prevalence of malocclusion, nasal septum deviation, arched palate, oral breathing, adenotonsillar hypertrophy, and short lingual frenulum than children with negative SCR (Tables 1 and 2). No differences in age, sex distribution, obesity, and IOPI measurements were identified between the 2 groups of participants.

### *3.2. Risk of positive SCR in children with short lingual frenulum*

A short lingual frenulum was found in 114 children (22.6%). Children with a short lingual frenulum were at significantly higher risk for a positive SCR compared to those with a frenulum of normal length when other factors possibly affecting the risk of OSAS (age, sex, tongue strength, obesity) were taken into consideration (Table 3).

## 4. Discussion

It was demonstrated that a short lingual frenulum is associated with increased risk of SDB as expressed by positive SCR in a school-age population, after considering the effect of other confounding factors such as age and obesity. This was an epidemiological, population-based study assessing the prevalence of and risk factors for SDB in children, and the potential role of short lingual frenulum in the pathogenesis of nocturnal upper airway obstruction was 1 of the study hypotheses tested.

A short lingual frenulum modifies the position of the tongue and impairs orofacial development, particularly in early life. More specifically, it has been associated with difficulties in sucking, swallowing, and speech [20,21]. Recent evidence indicates that a short lingual frenulum could lead to oral−facial dysmorphism. Orofacial changes have been shown to lead to mouth breathing, with modification of the position of the tongue, and secondary orthodontic abnormalities such as anterior and posterior crossbite, disproportionate growth of the mandible, and an abnormal growth of the maxilla. All of these anatomical changes affect the size of the upper airway and increase the risk of pharyngeal collapse during sleep [1,22].

Genetic factors and the arrangement of soft tissues in the orofacial region have been suggested as etiological factors in malocclusion [23]. The primary function of the lingual frenulum is to keep a balance between the growing bones and the tongue and the lip musculature during fetal development and to limit the movement of the muscular tissues such as lip, tongue, and cheeks [24]. The growth of soft tissues has a strong influence on the growth of hard tissues [24,25]. The tongue is such a soft tissue component that can affect the growth of the maxilla and mandible [24,26]. The equilibrium between the tongue and buccinator muscles is responsible for the development of normal arch width of the maxillary and mandibular arches [27]. The size, position, structure, and function of the tongue also have a potential role in the etiology of malocclusion [24].

A few researchers have reported on the relationship between malocclusion and a short lingual frenulum [24−28]. Defabianis et al proposed that ankyloglossia limits the upward movement of the tongue, thus preventing the formation of lip seal during swallowing and causing tongue thrusting and open bite [28]. Furthermore, the upward movement of the tongue is necessary to achieve normal width of the hard palate. Inability to lift the tongue upward results in unrestricted activity of the buccinator muscles and constriction of the maxillary arch, reduced maxillary intermolar and intercanine width, as well as reduced mandibular intercanine width [24,28]. Defabianis et al demonstrated that maxillary arch constriction, maxillary protrusion, crowding, and open bite were more common in subjects with short lingual frenulum compared to deep bite and spacing [23,28].

Higher nasal resistance and oral breathing in children with SDB result in an abnormal tongue position at rest and during sleep, which in turn reduces tongue movement and probably induces tongue hypotonia [14,18]. These factors are associated with skeletal malocclusions, and when the tongue is not in its normal position (ie, lying against the maxillary incisors and hard palate) but sits on the floor of the mouth, the modeling role played by the tongue on the oral cavity, upon every deglutition, is reduced [25,29−32].

In a previous study, we showed that children with SDB had lower tongue strength, as measured by the IOPI, than healthy children, a finding that was not reproduced in the present investigation [18]. Hence, measurement of tongue strength did not provide us with additional insight into the pathophysiological role of a short lingual frenulum. However, in the previous study, diagnosis of SDB was based on a different method, namely polysomnography.

In addition to Kotlow's method for recognizing a short lingual frenulum, other tools have been described. The Hazelbaker Assessment Tool for Lingual Frenulum Function has been used for infants, and Marchesan et al reported on the relationship between maximal interincisal mouth opening to tongue tip movement toward the incisal papilla [33−36]. Yoon et al validated the previous tools in a large cohort including participants 6−70 years of age [37].

## 5. Conclusion

In conclusion, in the present study, an association of short lingual frenulum with SDB, as expressed by a positive SCR, has been demonstrated. A multidisciplinary approach is necessary to allow early detection and timely treatment of craniofacial changes and potentially prevent SDB.

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**Table 1**

Abnormalities contributing to upper airway obstruction that are included in the Sleep Clinical Record (SCR).

| | Negative SCR (n = 462) | Positive SCR (n = 42) | *P* |
|---|---|---|---|
| Malocclusion | 207 (44.8%) | 37 (88.1%) | <0.001 |
| Nasal septum deviation | 35 (7.5%) | 10 (23.8%) | <0.001 |
| Arched palate | 230 (49.7%) | 40 (95.2%) | <0.001 |
| Oral breathing | 47 (10.2%) | 26 (61.9%) | <0.001 |
| Adenotonsillar hypertrophy | 79 (17.1%) | 18 (42.8%) | <0.001 |

**Table 2**

Additional characteristics potentially affecting the risk of sleep-disordered breathing that are not included in the Sleep Clinical Record (SCR).

| | Negative SCR (n = 462) | Positive SCR (n = 42) | *P* |
|---|---|---|---|
| Age, y | 10.1 ± 2.0 | 10.2 ± 3.1 | NS |
| Sex, male (%) | 199 (43.0%) | 23 (54.7%) | NS |
| BMI percentile | 67.4 ± 32.5 | 61.1 ± 35.5 | NS |
| Obesity, % | 64 (13.9) | 8 (18.1) | NS |
| Tongue strength, KPa | 46.4 ± 13.5 | 44.9 ± 12.8 | NS |
| Tongue endurance, sec | 14.5 ± 6.6 | 15.2 ± 8.2 | NS |
| Frenulum length,[a] mm | 18.5 ± 4.4 | 18.6 ± 4.0 | NS |
| Short lingual frenulum (%) | 96 (20.8%) | 18 (42.8%) | <0.002 |

BMI, body mass index; NS, not significant.
[a]Kotlow measurement.

**Table 3**
Multivariate logistic regression model assessing the association between a short lingual frenulum and risk of sleep-disordered breathing (positive Sleep Clinical Record) after adjustment for age, sex, tongue strength, and presence of obesity.

| Dependent variable: positive Sleep Clinical Record | B | SE | *P* | Odds ratio | 95% Confidence interval | |
|---|---|---|---|---|---|---|
| | | | | | Lower | Upper |
| Sex | −0.620 | 0.438 | 0.157 | 0.538 | 0.228 | 1.268 |
| Age | −0.023 | 0.112 | 0.834 | 0.977 | 0.784 | 1.216 |
| Obesity | 0.092 | 0.601 | 0.878 | 1.097 | 0.338 | 3.559 |
| Mean tongue strength (measured by IOPI) | −0.009 | 0.018 | 0.625 | 0.991 | 0.957 | 1.027 |
| Short lingual frenulum | 1.092 | 0.436 | 0.012 | 2.980 | 1.269 | 6.997 |
| Constant | −1.887 | 0.956 | 0.049 | 0.152 | | |

IOPI, Iowa Oral Performance Instrument; SE, standard error.

**Highlights**

- Recent evidence has focused attention on the presence of a short lingual frenulum in children with sleep-disordered breathing (SDB).
- In the present study, an association of short lingual frenulum with SDB, as expressed by a positive Sleep Clinical Record, has been demonstrated.
- A multidisciplinary approach is necessary to allow early detection and timely treatment of craniofacial changes and potentially to prevent SDB.