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Short lingual frenulum and head-forward posture in children with the risk of obstructive sleep apnea

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ABSTRACT

Background: Recent studies have shown that a short lingual frenulum is a potential risk factor for obstructive sleep apnea syndrome (OSAS) in children. A short frenulum leads to abnormal orofacial development and may consequently contribute to sleep-disordered breathing by narrowing the upper airways and increasing the risk of upper-airway collapsibility. The aim of this study was to assess the impact of a short lingual frenulum on the risk of OSAS in children.

Methods: Children from pre-, primary, secondary, and high school, aged 3–17 years, were included in the study. Parents/guardians were asked to fill in the Pediatric Sleep Questionnaire (PSQ), and then, children at risk of OSAS were enrolled in the study group. A control group was established randomly from patients with negative PSQ results. A physical examination, including measurements of head-forward posture (HFP) and length of the free tongue, inter-incisor distance and subjective high-arched palate evaluation was performed in children from both groups.

Results: A total of 1,500 PSQ questionnaires were distributed, and less than half (713) were returned correctly filled in. In the second part of the study, 135 children were evaluated: 67 in the study group and 68 in the control group. The mean ages were 9.4 ± 3.0 and 9.5 ± 3.1 years, respectively. Children in the study group had significantly shorter lingual frenula, higher HFP measures, and had a higher prevalence of a high-arched palate. Based on statistical analysis, a short lingual frenulum (OR 5.02 [1.58–15.94]).

Conclusions: 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. Furthermore, OSAS was associated with higher HFP, but no relationship was found between the two parameters.

1. Introduction

Obstructive sleep apnea syndrome (OSAS) in children is a form of sleep-disordered breathing that is characterized by partial or complete upper-airway obstruction and may lead to hypoxemia and hypercapnia. OSAS occurs in 3–10% of children, with a peak prevalence between 2 and 8 years of age [1]. Symptoms include snoring, gasping, sleep enuresis, witnessed apneas, and morning headaches. OSAS in children may lead to serious neurobehavioral problems, such as hyperactivity, daytime sleepiness, attention disorders, deterioration of cognitive functions, and learning difficulties [2,3].

Various risk factors of OSAS in children, including obesity, adenoid and tonsil hypertrophy, allergic rhinitis, and septal deviation, have been described [4]. Recent studies have shown that a short lingual frenulum may also contribute to the development of OSAS in children [5–7]. The shortening of the tongue frenulum, known as ankyloglossia, is an anatomic condition caused by an abnormally short or altered attachment of the lingual frenulum, which leads to movement restriction of the tongue [8]. It is associated with impaired suckling, chewing, and swallowing and may frequently lead to speech disorders [9–11]. Moreover, ankyloglossia has a negative impact on orofacial development leading to anterior and posterior crossbite, abnormal growth of the maxilla, and

Abbreviations: HFP, head-forward posture; OSAS, obstructive sleep apnea syndrome; PSQ, Pediatric Sleep Questionnaire; SCR, sleep clinical record.

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disproportionate growth of the mandible [6,12,13]. These anatomical changes can affect respiratory function by reducing the upper-airway size [14] and increasing the risk of upper-airway collapsibility during sleep [15].

Additionally, it has been reported that a short lingual frenulum also contributes to incorrect head positioning (forward extension of the head) [16], which might lead to a predisposition for OSAS [17,18]. The tongue has many myofascial and muscular attachments to the cervical muscles. The reduction of tongue mobility due to ankyloglossia requires the use of additional muscles for chewing, swallowing, and breathing. This hyperactivity of the cervical muscles causes their shortening and induces forward head posture [16,19]. At the 2018 Academy of Applied Myofunctional Sciences Conference, Fung [17] presented the results of a study carried out among children in Hong Kong and showed the relationships between ankyloglossia, head-forward posture (HFP), and obstruction of the upper respiratory tract.

PSG is the gold standard in the diagnosis of sleep apnea, however, it is an expensive test, hardly accessible and stressful for children, therefore we stand by the position that these tests should be performed only in children with the risk found on the basis of the questionnaires. Thus we decided to write an article in children with the risk of OSAS based on questionnaire and facing the unavailability of pediatric PSG.

The aim of this study was to assess the impact of short lingual frenulum on the risk of OSAS in children. Furthermore, it was intended to determine correlations between short lingual frenulum and HFP.

2. Materials and Methods

Children from preschools, primary schools, secondary schools, and one high school, aged 4–17 years, were enrolled in the study. All children in the study were Caucasian race (inhabiting the rural area of Mazovian district where the study was conducted). In the first part of the study, 713 children participated, with 347 (49%) boys and 366 (51%) girls. The mean age was 9.8 years. During the second part of the study, 135 children were evaluated, 67 of whom were in the study group and 68 in the control group. The mean ages were 9.4 ± 3.0 and 9.5 ± 3.1 years, respectively. Of the studied children in the second part, 51% (69) were girls and 49% (66) were boys.

The cross-sectional study was conducted between September 2018 and March 2019 in rural areas of central Poland. Bioethical Committee approval was obtained (AKBE/207/2018).

The study consisted of two parts. First, a screening survey was performed using the Pediatric Sleep Questionnaire (PSQ) for OSAS in children by distributing questionnaires in schools. Second, the children at risk of OSAS were asked to participate in a medical examination and were assigned to the study group. Additionally, a control group was established randomly from children not presenting OSAS risk (matched by sex and age), and they underwent the same procedures as the study

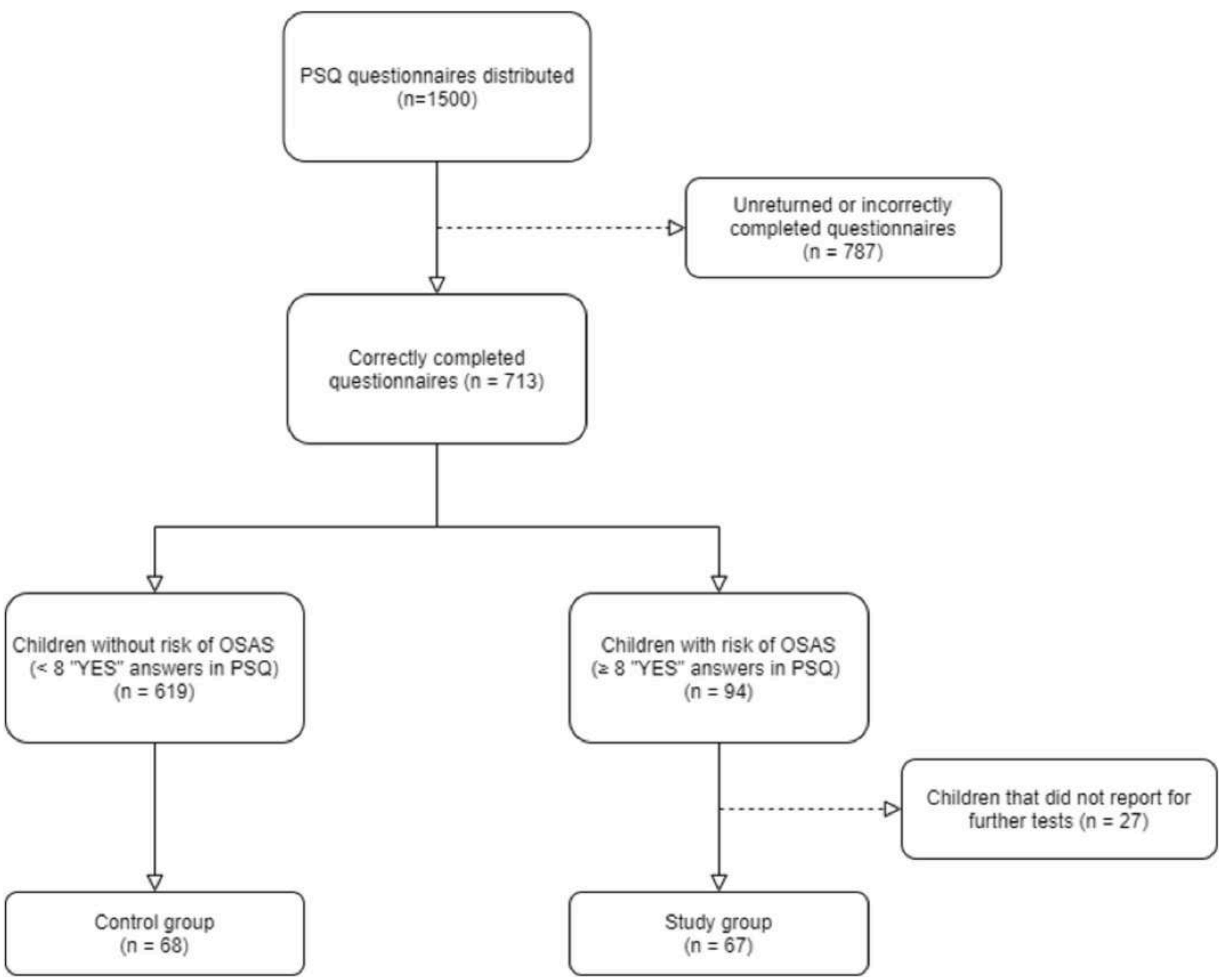

Fig. 1. Flowchart demonstrating the selection of the study and control groups.

group. The study scheme is presented in Fig. 1.

2.1. Screening of children with the risk of OSAS

The screening questionnaires were distributed by teachers in preschools, primary schools, secondary schools, and one high school. After being filled out by the parents/caregivers, the survey was returned in a sealed envelope to the teacher by the students. Approximately 1,500 questionnaires were distributed, of which 713 were correctly completed and returned.

The PSQ was used to conduct the study. The PSQ is characterized by high specificity and sensitivity in detecting OSAS risk in children and does not require the involvement of medical staff. The current guidelines of the Working Group of the European Respiratory Society recommend it as a screening tool [20,20,21,21]. Moreover, the specificity of the PSQ is the highest of all available questionnaires for children, thus providing the lowest percentage of false positive results. The authors of the PSQ recommend that 8 or more "YES" answers in the questionnaire qualify a child to be in the OSAS risk group [2,2,22,22]. Children who received 8 or more points (8 or more affirmative answers) in the PSQ were enrolled in the study group (n = 94). Sixty-seven children out of 94 entered the second part of the study, which was carried out in an outpatient setting and consisted of a medical history and physical examination. Afterwards, the control group was randomly selected from the group of children without the risk of OSAS (those who received less than 8 points in the PSQ). Both groups were matched by sex and age.

2.2. Medical examination

The second part of the study consisted of a physical examination and medical history. Children from both the study and control groups participated in this part of the study.

The physical examination of the child included measurements of height and weight, BMI, BMI percentile, assessment of the oral cavity for the presence of a high-arched palate, and evaluation of anthropometric parameters, such as HFP and length of the free tongue,inter-incisor distance - the method of measurement is described further. The high arched palate was examined by one person and only with the subjective method of evaluation (YES/NO). High-arched palate was evaluated according to its curvature - as an abnormally pronounced curvature angled superiorly along the palatal midline.

2.2.1. Measurement of head-forward posture (HFP)

To measure HFP, the patient was asked to stand against the wall, slightly touching it with his/her back, and look straight ahead. The distance between the wall and the furthest point of the cervical lordosis was measured, as shown in Fig. 2.

2.2.2. Evaluation of length of the free tongue

The tongue frenulum was measured using the Quick Tongue-Tie Assessment Tool (Fig. 3), which measures the inter-incisor distance and length of the free tongue. Inter-incisor distance is the distance between the upper and lower incisors, measured with the mouth wide open, as shown in Fig. 4. The length of the free tongue is the distance between the insertion of the lingual frenulum into the base of the tongue and the tip of the tongue, measured as shown in Fig. 5. If the length of the free tongue is $\leq$ 16 mm, the lingual frenulum is considered short [23].

2.3. Statistical analysis

SAS 9.4 software was used to assess the variables studied. The student's t-test for independent groups and the chi-square test (χ^2 test) were used to assess the significance of differences. Interdependence of variables was evaluated by Pearson's correlation test. The statistical significance of a p-value < 0.05 was assumed.

3. Results

The study group (PSQ≥8) included 67 children and the control group (PSQ<8) - 68 children (total 135 children). The total group characteristics are shown in Table 1. The groups did not differ in terms of height, weight, and inter-incisor distance. The results such as BMI, BMI percentile, length of the free tongue and HFP are presented in three age groups: 3–7, 8–12, 13–16 in Figs. 6 and 7. The number of children in each age group was: 36 (3–7), 72 (8–12) and 27 (13–16).

Children from the study group had significantly shorter lingual frenula (determined by the length of the free tongue [23]), higher HFP measurements, and higher prevalence of high-arched palate compared to the control group (Table 1).

A short tongue frenulum was found in 20 children, 16 of whom were in the study group. In a group of children with short frenula, the risk of positive PSQ was 5 times higher than in a group with normal frenulum length (OR 5.02 [1.58–15.94]). The correlation between the length of the free tongue and HFP was evaluated using Pearson's correlation test, but no relationship was found (r = 0.058; p = 0.507) (Table 2).

4. Discussion

The PubMed, Medline, and Cochrane databases were searched using the following terms: "ankyloglossia," "tongue-tie," "short lingual frenulum," and "obstructive sleep apnea." Three studies and one guideline

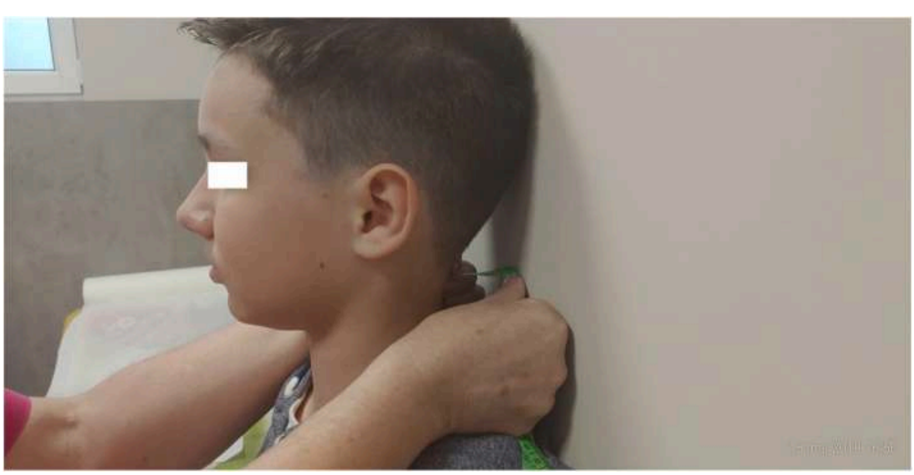

Fig. 2. Measurement of HFP.

Fig. 3. Quick tongue-tie assessment tool.

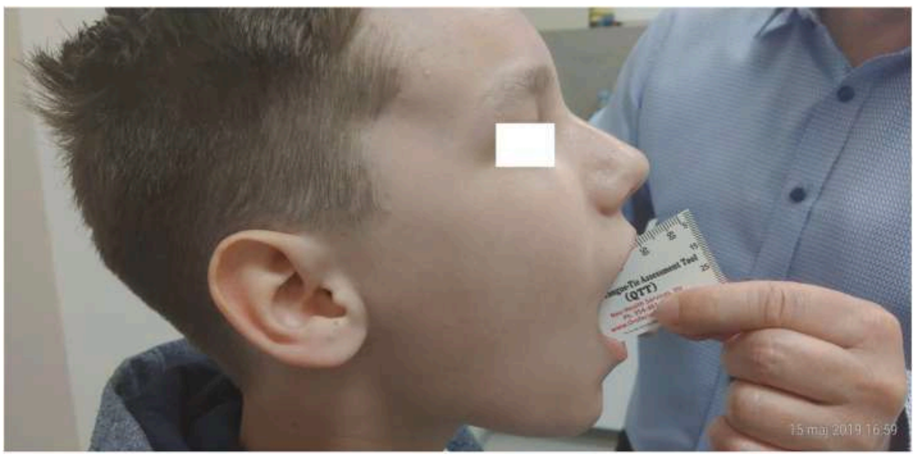

Fig. 4. Measurement of the inter-incisor distance.

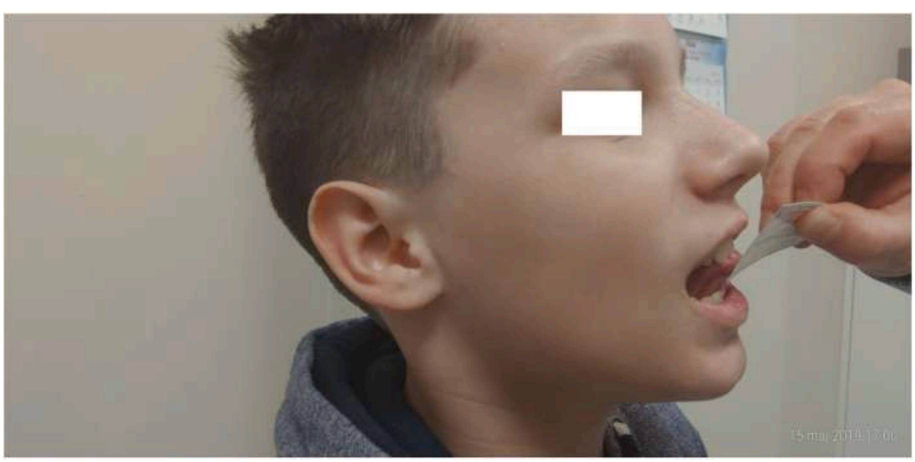

Fig. 5. Measurement of the length of the free tongue.

assessing the relationship between a short lingual frenulum and OSAS in children were found.

Huang et al. [6] conducted a retrospective study to identify short lingual frenula within children with suspected OSAS. In all 27 children participating in the study, changes in the orofacial anatomy and reduced oral cavity size were identified. All participants were unaware of the abnormal length of the lingual frenulum. The majority of the patients underwent treatment via adenotonsillectomy, frenectomy, or orthodontic intervention. The authors concluded that an assessment of the frenulum length should be performed in every child with OSAS, as should screening for OSAS in every child with a short frenulum. Additionally, a frenectomy should be performed as early as possible to prevent abnormal orofacial development, which can consequently lead to sleep apnea [6].

Guilleminault et al. [5] investigated the relationship between a short lingual frenulum and OSAS in a group of 150 children with OSAS, 63 of whom had short lingual frenula. Among children with ankyloglossia, mouth breathing and abnormal anatomical findings were observed more frequently. Moreover, in 60 cases, at least one family member had a short lingual frenulum and a positive diagnosis of sleep-disordered breathing. The authors suggested that children with a short frenulum should be screened for OSAS, as it is a risk factor for the development of sleep-disordered breathing.

Villa et al. [7] evaluated 504 children, 42 of whom were at high risk

Table 1
Characteristics of the study and control groups.

	Study group (n = 67) (PSQ ≥ 8)	Control group (n = 68) (PSQ < 8)	p-value
Age, y	9.4 ± 3.0	9.5 ± 3.1	NS
Sex, % female (n)	51% (34)	51% (35)	NS
Height, cm	139.6 ± 18.7	141.2 ± 19.0	NS
Weight, kg	37.9 ± 15.9	38.5 ± 15.9	NS
HFP, mm	77.2 ± 20.0	70.2 ± 18.1	0.0306
Inter-incisor distance, mm	46.6 ± 6.1	46.3 ± 5.8	NS
Length of the free tongue, mm	21.9 ± 6.0	24.1 ± 4.9	0.0194
Short lingual frenulum[a], n (%)	16 (24%)	4 (6%)	0.0033
High arched palate, n (%)	27 (40%)	4 (6%)	<0.001

[a] Length of the free tongue ≤ 16 mm, NS—not significant.

for OSAS based on the Sleep Clinical Record (SCR) evaluation. The study did not show any differences in mean frenulum length between the SCR positive (at risk of OSAS) and negative (without risk of OSAS) groups (frenulum length 18.6 ± 4.0 and 18.5 ± 4.4, respectively). Children with a short lingual frenulum were at significantly higher risk for a positive SCR compared to those with a normal frenulum length (OR 2.98 [1.269–6.997]; p = 0.012). The study showed that a short frenulum is more prevalent in children with a risk of OSAS and that it is a risk factor for sleep apnea.

In all the aforementioned studies, the relationships between ankyloglossia and OSAS were described. However, according to the guidelines in "Clinical Consensus Statement: Ankyloglossia in Children" published in April 2020 [24], which took into account only one study, there is not enough evidence that a short lingual frenulum is a risk factor of OSAS.

In our study, the mean length of the free tongue was 23.0 ± 5.6 for the entire group and 21.9 ± 6.0 and 24.1 ± 4.9 for the study and control groups, respectively, which is higher than that reported in the literature. In the above mentioned study carried out by Villa et al. [7], the average length of the free tongue for the groups with and without the risk of OSAS was 18.6 ± 4.0 and 18.5 ± 4.4, respectively. In a study carried out by Yoon et al. [25,26] on 1,052 patients, aged 6–70 years, who reported for an orthodontic evaluation, the mean length of the free tongue was 17.5 ± 5.5 for the total group and 16.3 ± 4.4 and 16.9 ± 5.2 for the subgroups of children aged 6–11 years and 12–17 years, respectively. The characteristics of the studies are presented in Table 3. The differences may stem from the selection of groups for the survey and the small size of our group. More studies are needed to determine the average length of the free tongue in the general pediatric population.

In the current study, we found that children with a risk of OSAS based on the PSQ had a shorter length of the free tongue, which is a measurement used to determine a short lingual frenulum. Additionally, children in the study group were more likely to have a high-arched palate. A high-arched palate is a known risk factor for sleep apnea [27–29]. By restricting the correct movements of the tongue and resulting in its incorrect positioning, a short lingual frenulum contributes to the abnormal growth of the maxilla and development of a high-arched palate [6,29]. In a study carried out by Guilleminault et al. [5], children with short lingual frenula had a higher prevalence of high-arched palates compared to the control group (80% vs. 8.75%, respectively; p < 0.0001). In the study conducted by Villa et al. [7], 95.2% of children at risk of OSAS had high-arched palates, compared with patients without the risk of sleep apnea (p < 0.001). Shafiek et al.

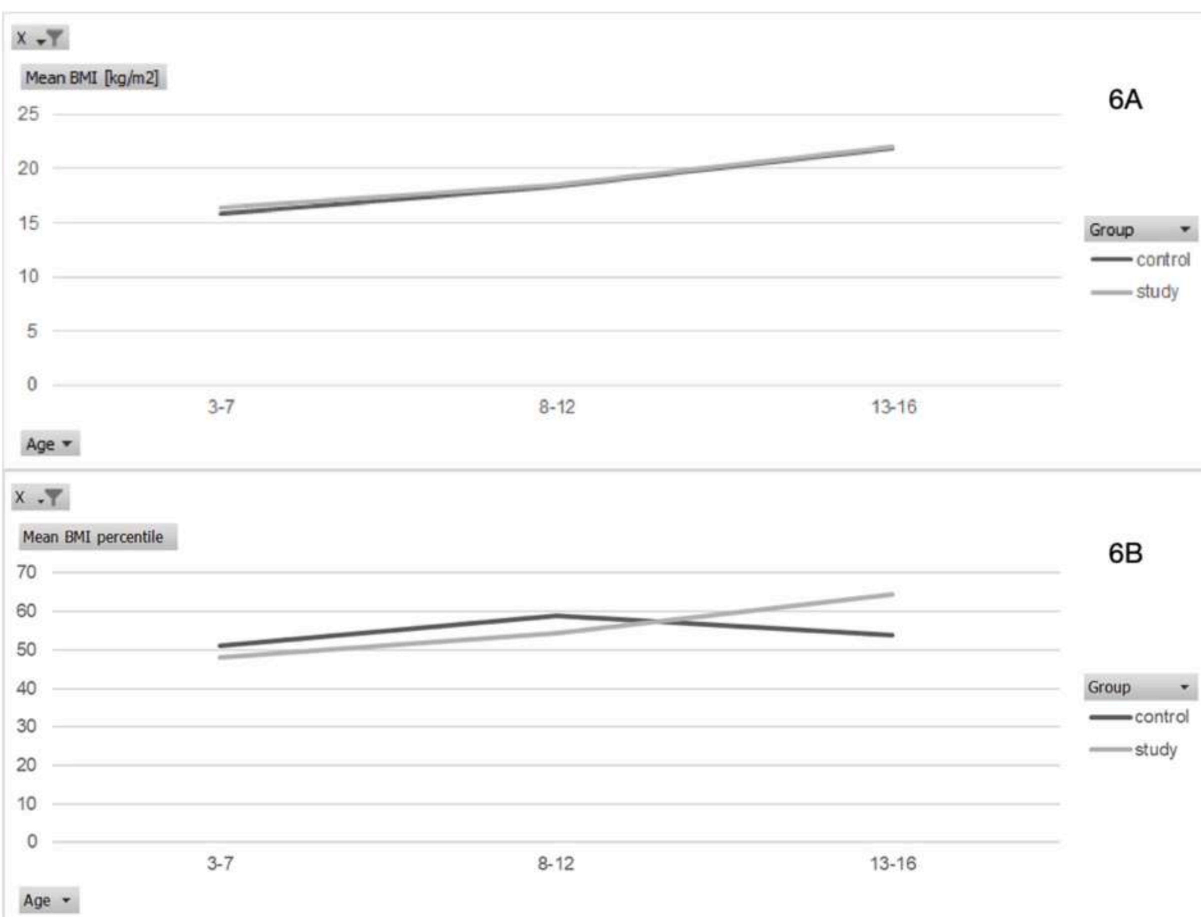

Fig. 6. A,B. Mean BMI (A) and mean BMI percentile (B) in age groups.

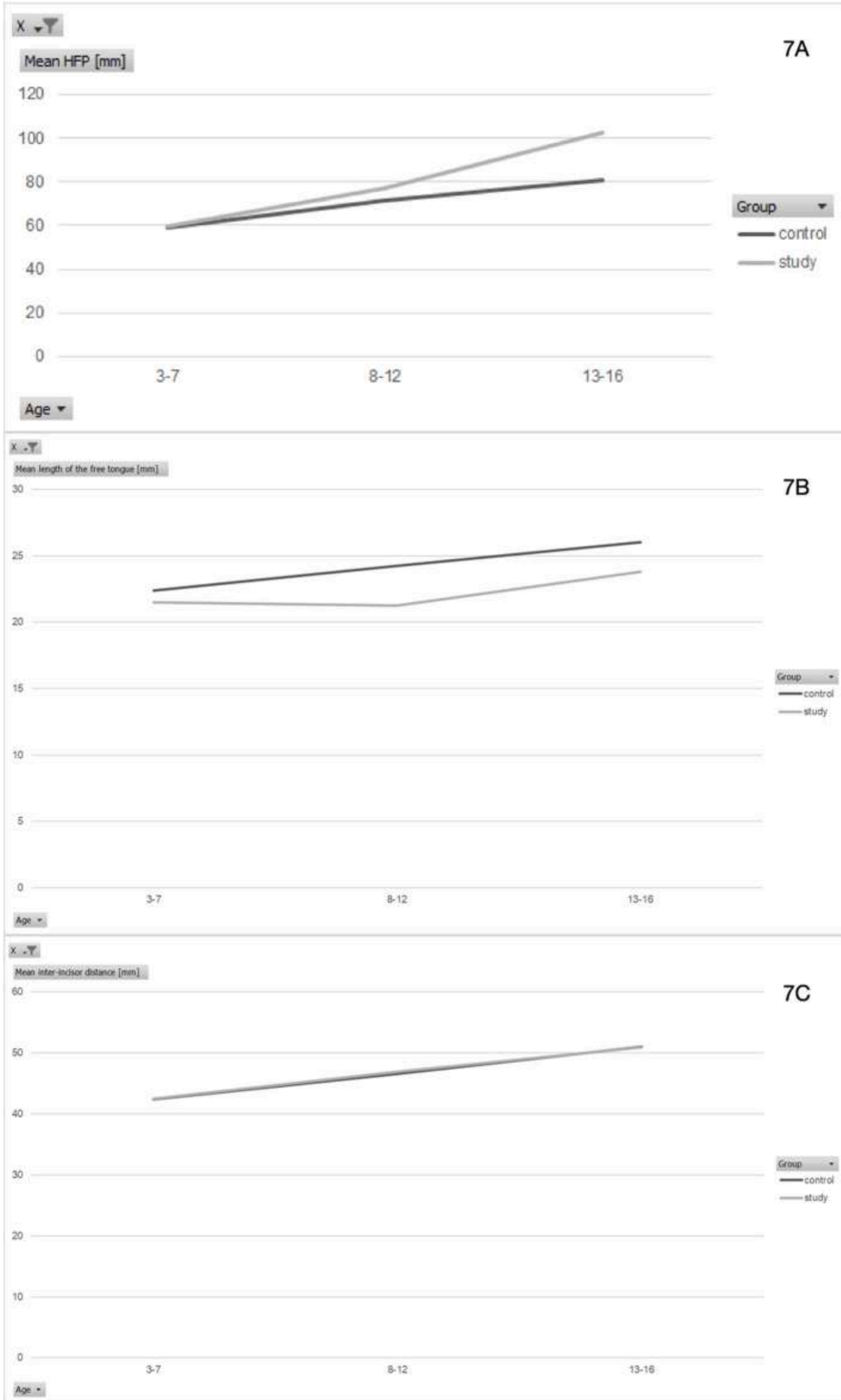

Fig. 7. A–C Mean HFP (Head Forward Position) (A), mean length of the free tongue (B), mean inter-incisor distance (C) in three age groups.

[30] evaluated the prevalence of OSAS in children presenting with nocturnal enuresis. This study showed that, in a group of children with OSAS, 66.7% had high-arched palates, compared to 26.7% in a non-OSAS group ($p = 0.01$).

In our study, the mean inter-incisor distance was 46.6 ± 6.1 for the study group and 46.3 ± 5.8 for the control group. There are no definite norms for the inter-incisor distance in the literature. This measure depends on the age, height, gender and race of the child. In a study conducted on 1059 Turkish children, the maximum inter-incisor distance was 33.24 ± 5.54 for females; 33.32 ± 5.71 for males (28.63 ± 4.34 for 3–5 years; 33.52 ± 4.84 for 6–11 years; 37.35 ± 5.52 for 12–15 years) [31]. According to another study carried out on 602 Indian children, the mean inter-incisor dimension in the group of the youngest children (4–5 years) was 41.34 ± 4.14, increasing with age to 51.73 ± 6.46 in the age group of 14–15 years [32]. While in the study performed on more than 20,000 Swiss children, the mean inter-incisor distance was

Table 2
Pearson's correlations between length of the free tongue and head-forward posture (HFP).

		R (X,Y)	r2	t	p
All children		0.0576	0.00332	0.666	0.507
Length of the free tongue	23.0 ± 5.6				
HFP	73.7 ± 19.3				
Study group		0.111	0.0123	0.900	0.371
Length of the free tongue	21.9 ± 4.9				
HFP	77.2 ± 20.0				
Control group		0.0907	0.00823	0.740	0.462
Length of the free tongue	24.1 ± 6.0				
HFP	70.2 ± 18.1				

Table 3
Studies evaluating frenulum (determined as the length of the free tongue).

Study	Age, y	The length of the free tongue[a], mm
Yoon et al. [25,26]	6–11	16.3 ± 4.4
	12–17	16.9 ± 5.2
Villa et al. [7]	6–14	Children at risk of OSAS: 18.6 ± 4.0
		Children without risk of OSAS: 18.5 ± 4.4
Our results	4–17	Total group: 22.0 ± 5.6
		Children at risk of OSAS: 21.9 ± 6.0
		Children without risk of OSAS: 24.1 ± 4.9

[a] Kotlow measurement [23].

45 mm for both boys and girls. In the cited study, the authors proposed percentile norms depending on the age and gender of the child [33]. Although there are no clear norms for the above-described parameter, authors of the studies agree that it increases with age and height of children.

Moreover, patients from the study group in our research had significantly higher HFP measurements than the controls. Studies have found that correct head posture is an important factor for proper breathing. In a study carried out by Piccin et al. [18], head hyperextension and anteriorization were correlated with the severity of OSAS. Kim et al. [34] found that HFP was negatively associated with respiratory functions and positively correlated with accessory muscle activation.

A short lingual frenulum has also been reported to be a possible risk factor leading to abnormal body posture. Olivi et al. [16], in a study published in 2012, described the relationship between ankyloglossia and body posture. Ankyloglossia is associated with a higher and more advanced position of the hyoid bone, which results in an increased tone of extrinsic and suprahyoid lingual muscles. This leads to posture abnormalities, including patients leaning forward with their head and shoulders protruding; consequently, cervical hyperlordosis and dorsal kyphosis may occur [16]. However, in our study, we did not find a correlation between the length of the free tongue and HFP. More studies are needed to assess this relationship.

Ankyloglossia limits the upward movement of the tongue, thus preventing the formation of lip seal during swallowing, leading to tongue thrusting, which in turn can cause open bite. The upward movement of the tongue is necessary for the creation of normal width of the hard palate, therefore anykologlossia results in the reduction of maxillary intermolar and intercanine widths. Additionally, pulling of short lingual frenulum resulting in constriction of mandibular anterior region may lead to reduction of mandibular intercanine width [8,13]. Abnormal bone growth stimulation, an absence of nasal breathing (due to anatomical and muscle tone dysfunctions) with secondary development of mouth breathing are responsible for the abnormal development of the oral-facial bone structures supporting the upper airway, thus increasing the risk of UA collapse during sleep [35].

The only sleep registry we performed at the time of study was night pulse-oximetry, but it did not let us draw any conclusions, what was discussed already by many authors. Thus we decided to write an article in children with the risk of OSAS based on questionnaire and facing the unavailability of pediatric PSG.

5. Conclusions

In conclusion, the conducted study identified a relationship between a short lingual frenulum and the risk of OSAS in children (expressed as a positive PSQ). Detecting and addressing ankyloglossia in children is necessary before it leads to orofacial changes, malocclusion, and consequently, sleep apnea. Furthermore, OSAS was associated with higher HFP, but no relationship was found between the two parameters.

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Declaration of competing interest

Marcin Burghard - no conflict of interest.
Eliza Brożek-Mądry - receives grant-funded research support from Probios (probiotic company).
Janusz Sierdziński - no conflict of interest.
Antoni Krzeski - no conflict of interest.

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