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Association of orofacial dysfunction and sleep disordered breathing among Indian primary school children

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ABSTRACT

Introduction: Sleep-disordered breathing (SDB) ranges from partial obstruction of the upper airway resulting in snoring to total upper airway obstruction leading to obstructive sleep apnea. The impairment in the dynamics of the stomatognathic system is termed as orofacial dysfunction. This study investigates the prevalence of orofacial dysfunction and sleep-disordered breathing in primary school children and identifies their correlation.
Methods: A total of 560 forms were distributed to 8 primary schools in Belagavi city. Among them, 482 parents responded (86% response rate), which included 239 boys (49.58%) and 243 girls (50.41%). All the participants were screened for orofacial dysfunction using Nordic Orofacial Dysfunction Test-screening (NOT-S) and sleep-disordered breathing using the Pediatric Sleep Questionnaire (PSQ).
Result: A positive direct correlation of sleep-disordered breathing with orofacial dysfunction ($r = 0.47$; $p \leq 0.001$) was noted. A total of 41(8.58%) children were found to be at risk of sleep-disordered breathing with a score less than or equal to eight, based on (PSQ) Pediatric Sleep Questionnaire, and 156 (32.6%) children showed symptoms of orofacial dysfunction based on Nordic Orofacial Test–Screening (NOT-S).
Conclusion: The study demonstrates that around 32.6% of children had orofacial dysfunction symptoms, and 8.58% of children were at risk for sleep-disordered breathing, girls having a greater risk as compared to boys. There was a positive correlation between orofacial dysfunction and sleep-disordered breathing among children aged 6–12 years.

1. Introduction

Sleep-disordered breathing (SDB) ranges from partial obstruction of the upper airway resulting in snoring to total upper airway obstruction leading to obstructive sleep apnea.[1] Three elements contribute to sleep-disordered breathing: anatomical structure, neuromotor tone, and inflammation.[2] The critical consequence of pathophysiologies of these elements is upper airway obstruction. Upper airway obstruction during sleep is associated with loud snoring, arousal, sleep fragmentation, intermittent hypoxemia and hypercapnia, nocturnal hypertension, daytime sleepiness, deterioration in academic performance and cognitive abilities.[3] Sleep-disordered breathing showed a prevalence of 11.4%–47.5% in North India and 4.8% to 5% in South India.[4–6]

Literature suggests that snoring or upper airway obstruction during sleep is associated with craniofacial modification in children.[7] Craniofacial transformation and orofacial growth co-occur. The two centers for orofacial development are the intermaxillary synchondrosis and alveolodental ligament that are active at birth and remain so even after maximal orofacial growth.[4] The most rapid orofacial growth occurs between birth and two years of age. However, the growth remains operational until the child is six years of age. Around 60% of children gain almost permanent craniofacial structure by the age of 6 years.[5] The change in craniofacial structure sometimes causes impairment in orofacial function,[8] resulting in nasal obstruction and parafunctional habits such as mouth breathing, bruxism, posturing the tongue forward, incompetence of lip, open mouth rest posture, finger, and thumb sucking, and lips, tongue, fingers, and cheeks biting, etc.[6] The orofacial function is a phenomenon in which the central nervous system, neuromuscular system, and stomatognathic system work simultaneously.[7] The impairment in the dynamics of the stomatognathic system is termed as

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orofacial dysfunction.[9]

Sustained daytime hypertension, increased cardiovascular and cerebrovascular morbidity, and mortality has been reported as long-term consequences of sleep-disordered breathing.[8] Various studies have evaluated the association between sleep-disordered breathing and orofacial dysfunction symptoms, which reports that there is a significant relationship between them.[1,10] However, the orofacial symptoms in the studies were evaluated either by using a questionnaire or using a tool that measures the physical characteristics of orofacial behavior. None of the studies have evaluated the primary motor and orofacial sensory function, including chewing, swallowing, and speech. Also, the tools used were developed to evaluate orofacial function in obstructive sleep apnea (OSA), which is the most severe form of sleep-disordered breathing. The mildest form of orofacial dysfunction, which has a high potential of progression later in life, is not yet evaluated.

In the Indian scenario, Orofacial dysfunction in children is the almost untouched topic of research to date. No studies regarding the correlation between orofacial dysfunction and sleep-disordered breathing have been conducted in India to the best of our knowledge. Furthermore, studies report a lack of awareness about the child's sleep health and breathing disorder among parents and health professionals in India.[11] This lack of awareness may lead to Indian children being undiagnosed and untreated. Therefore, this study investigates the prevalence of orofacial dysfunction and sleep-disordered breathing among primary school children in Belagavi and identifies the correlation between them during their craniofacial development.

2. Methods

A cross-sectional study among school children was conducted from December 2019 to March 2020 in Belagavi, Karnataka, India, after obtaining ethical approval from the Institutional Ethical Committee.

The sample size of 426 was arrived at with a statistical power calculation of 90% and 5% level of significance. A stratified random sampling technique was performed to ensure that the sample is representative of all primary schools of Belagavi. A number was allotted for all the primary schools from East, west, north, and south of Belagavi. Eight schools were selected using the randomization table method.

The principal investigator approached the school principals for permission to conduct the study, and the rationale of the study was explained. After acquiring their approval, they were provided with the consent forms to be distributed to the students in the age group of 6–12 years, i.e., children from first to the seventh standard. Ten students (5 male and five female) assuming a 20% non-response rate; from each standard, students were randomly selected using the lottery method from all the schools. Class teachers distributed the forms and requested the students to get them filled by their parents. In total, 560 forms were distributed to 8 primary schools in Belagavi city. Among them 482 parents responded (86% response rate); which included 239 boys (49.58%) and 243 girls (50.41%). The class teachers were then requested to fix an appointment with those parents who consented to their and their child's participation in the study. On the day of the examination, the parents were first interviewed by the research assistant seeking information; like if their child is diagnosed with a known developmental disability and craniofacial anomalies and whether they have any cough and cold symptoms. If the answer was "yes," the child was excluded from the study. Four children (one female and three male) were excluded from the study due to cold and cough symptoms on the day of examination. Thus, 478 children, 236 boys (49.4%), and 242 girls (50.6%) were included for further evaluation. NOT-S interview (Figure 1) and PSQ (Figure 2) were administered to the parents through an interview by the research assistant.[5] The principal investigator performed the evaluation of children for orofacial dysfunction using NOT-S examination subsequently on the same day.

3. Outcome measure

PSQ is a valid and reliable instrument with 0.88 and 0.86 validity and reliability, respectively. It is used to evaluate the symptoms of disordered sleep breathing. It has 22 items: snoring, breathing problems, mouth breathing, daytime sleepiness, and behavior problems. Each item has three response options; Yes, No, and don't know. Yes, response scores one, and no or don't know response scores zero. Children with eight or more scores were considered as high risk for SDB, whereas children with scores less than eight were considered as low-risk for SDB.[12]

NOT-S is a screening tool to identify the area of orofacial dysfunction. It is a freely available screening tool in various languages. It has two parts composed of a structured interview and clinical examination. The structured interview consists of six domains with verbal 'Yes' and 'No' response format. If the response to one or more items within a domain is 'yes,' the dysfunction criterion is fulfilled; if the response is 'No,' it means there is no dysfunction. The examination part also consists of six domains; each domain contains a variable number of items. 'Yes' in any item of the domain indicates dysfunction in the scored domain.[5]

4. Data analysis

The characteristics of participants and prevalence rates were described using descriptive statistics. A Chi-square cross-tabulation test was performed to assess the differences between Sleep-disordered breathing and or facial dysfunction in association with gender. Spearman correlation test was performed to evaluate the correlation between sleep-disordered breathing and orofacial dysfunction with age and correlation between (NOT-S) and (PSQ). The P-values of <0.05 were considered significant.

5. Results

Among 478 children recruited, 236 (49.4%) were boys, and 242 (50.6%) were girls with a mean age of 8.92 $\pm$ 2.05 years and an age range of 6–12 years (Table 1).

A total of 41(8.58%) children were found to be at risk of sleep-disordered breathing with a score less than or equal to eight, based on (PSQ) Pediatric Sleep Questionnaire, and 156 (32.6%) children showed symptoms of orofacial dysfunction based on Nordic Orofacial Test–-Screening (NOT-S). Among them, the number of children with at least one domain affected was 44, followed by breathing and nose breathing and nose breathing under which 28 children were affected, followed by sensory function and drooling under which 27 children were affected, and the least affected domain was facial expression under which one child was affected.

Chi-square cross-tabulation test on PSQ score and NOT-S scores with the gender revealed that there was a significant difference between PSQ score and gender ($\chi2 = 21.699$: $p = 0.027$). NOT-S and gender did not show significant difference ($\chi2 = 0.545$: $p = 0.969$) (Table 2). There is a

Table 1
Age and gender distribution.

Characteristics	n (%)
Gender	
Female	236(49.4)
Male	242(50.6)
Age	
6	79(16.5)
7	65(13.6)
8	72(15.1)
9	61(12.8)
10	65(13.6)
11	69(14.4)
12	67(14.0)

Table 2
Correlation between genders with sleep disordered breathing and orofacial dysfunction based on PSQS & NOTS Score.

Score		Gender		Chi Square value	p-value
		Male	Female		
PSQS	0	47	45	21.699	0.027*
	1	17	36		
	2	44	28		
	3	31	32		
	4	19	25		
	5	29	18		
	6	24	20		
	7	8	14		
	8	5	10		
	9	6	8		
	10	3	6		
	11	3	0		
NOTS	0	160	162	0.545	0.969
	1	43	42		
	2	20	21		
	3	12	16		
	4	1	1		

*Denotes statistically significant at $p < 0.05$.

negative indirect relationship between age and PSQ ($r = -0.136$: $p = 0.003$) and between age and NOT-S ($r = -0.133$: $p = 0.004$). There was a positive direct correlation of sleep-disordered breathing based on (PSQ) with orofacial dysfunction based on Nordic Orofacial Test–Screening ($r = 0.47$: $p = 0.001$) (Table 3).

6. Discussion

The present study used a valid and reliable PSQS with a 0.88 and 0.86 validity and reliability to evaluate sleep-disordered breathing symptoms and NOT-S to evaluate orofacial dysfunction among 478 children aged 6–12 years in Belagavi city.

In the present study, 32.6% of children reported orofacial dysfunction symptoms, among which the number of children with at least one domain affected was 44. The highest affected domain was 'habits,' under which 9.2% of children were seen. The number of children with the least affected domain ranges from 0.2% to 1.5%. This result is in accordance with the previous reports that revealed that the most commonly affected NOT-S domain in normal children was 'Habit.'[13,14]

Sleep-disordered breathing has previously been reported to be (51.1%) among preschool and school-aged children in rural India.[15] This rate is higher than reported in Dutch children (25%), Italian children (4.9%), Turkish children (7%), and urban Indian children (11.4%).[13,14,16] In this study, the prevalence of sleep-disordered breathing reported is 8.58%, which is less than that reported in rural Indian children, as this study was also conducted in the urban area. A higher prevalence of sleep-disordered symptoms among children in rural areas than in urban areas has been reported previously.[13]

The present study showed a higher risk of SDB among female children, with 58% being affected as compared to 41% of male children, which is in agreement with a previous study that reported a higher risk of sleep disorders in females than males.[17] The reason for this finding may be attributed to added descending mandibular growth, lower facial height, and anteriorly positioned hyoid bone in females than in males. Studies also show that these craniofacial features are more prevalent in severe sleep-disordered breathing.[18] However, some of the other studies reported a lower risk of sleep-disordered breathing in females than males.[19–21] This difference in the prevalence can also be attributed to gender bias that may influence the parents' reporting regarding snoring in boys and girls.[18]

The result also shows a positive direct association between sleep-disordered breathing and orofacial dysfunction among school children. This result agrees with Baidas et al., which concludes that there is a strong relationship between orofacial symptoms and sleep-disordered breathing and found that the history of digit sucking habit is associated with sleep-disordered breathing.[9] They have assessed orofacial dysfunction symptoms using a questionnaire that included questions like digit sucking habits, facial muscle pain, temporomandibular joint pain, and bruxism symptoms. In the present study, we have used a standardized tool for assessing orofacial dysfunction, which contains a wider orofacial function area. Our study result is also in agreement with the study by Huynh et al., which reported that the association between thumb/finger sucking histories is statistically significant with heavy breathing at night.[19]

Mouth breathing during the early stage of life impairs the temporomandibular joint,[21,22] and the prevalence of SDB is high in individuals with temporomandibular dysfunction.[23] The mild orofacial dysfunction symptoms can progress into severe obstructive sleep apnea.[24,25] In addition, the findings of this study suggest that majority of mild orofacial dysfunction symptoms remain unnoticed in the growing age of the children. This issue can be resolved by routine evaluation of orofacial functions in children and using various intervention strategies during the early life stages. Therefore, screening children for early identification of orofacial dysfunction is necessary to recognize and intervene the orofacial dysfunction and prevent its adverse consequences in later life.

The study did have some inherent limitations as the results for sleep-disordered breathing and orofacial dysfunction interviews were based on parents' opinions, which may be subjective to a certain extent. The accuracy of the results could be enhanced by the use of comprehensive and objective sleep-disordered breathing tools like polysomnography and contact devices. Furthermore, clinical evaluation of the children along with questionnaire may yield more objective information and take imminent studies to an advanced level, which we plan to undertake subsequently.

7. Conclusion

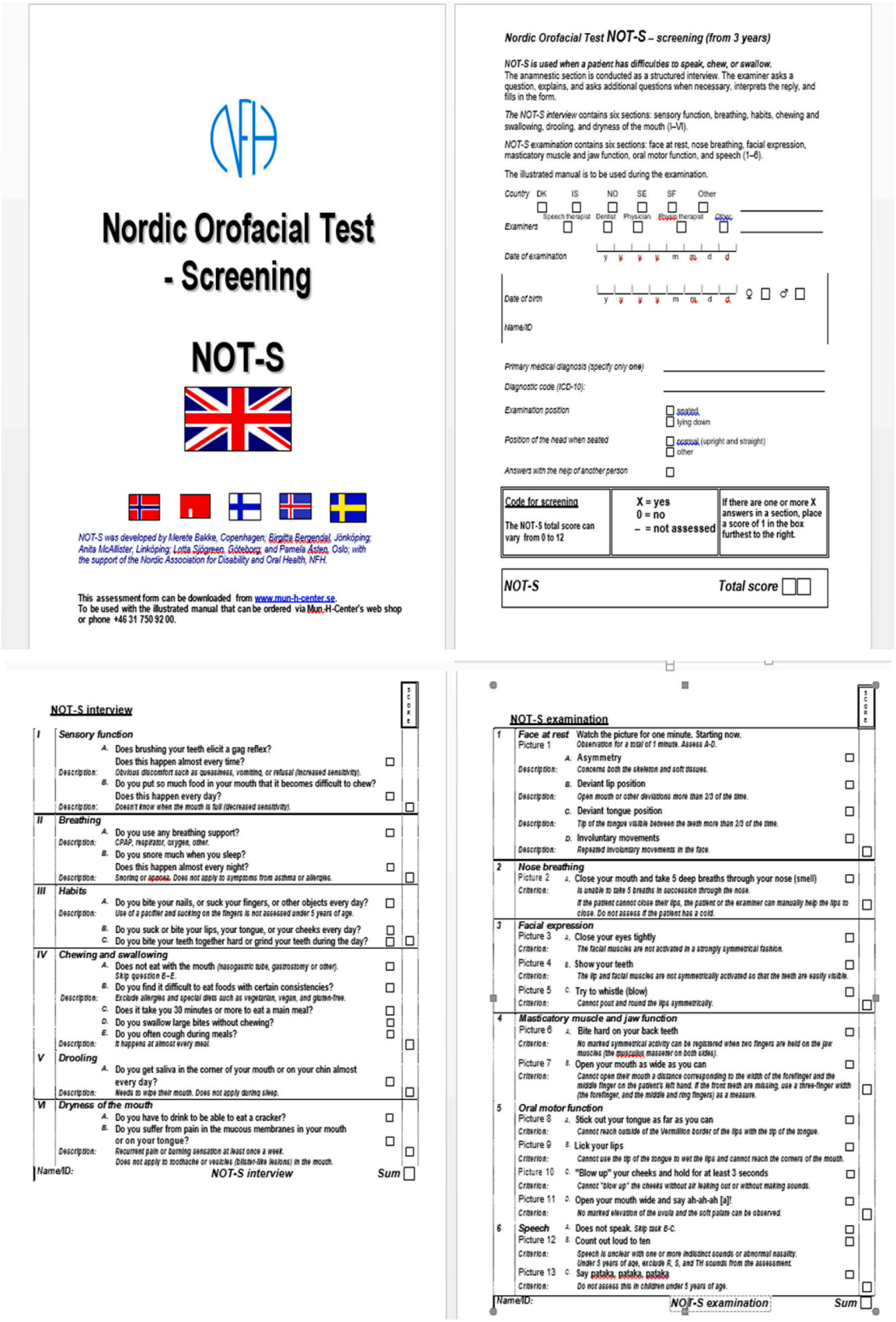
We found that 32.6% of children have symptoms of orofacial dysfunction, and 8.58%, children are at risk of sleep-disordered breathing, with girls being at higher risk than boys. There is a positive correlation between sleep-disordered breathing and orofacial dysfunction in the age group of 6–12 years. This study underscores the importance of screening children in the early stage of life for orofacial dysfunction and SDB, which is vital to prevent various adverse consequences during childhood and later in life. Further studies, including larger sample sizes with broader age groups and more comprehensive tools for sleep-disordered breathing, are required to generalize the result in a larger population.

Table 3
Association between Age and sleep disordered breathing, age and orofacial dysfunction and sleep disordered breathing & orofacial dysfunction based on PSQS & NOTS Score.

variables	r-value	p-value
Sleep disordered breathing and orofacial dysfunction	0.47[a]	0.001*
Age and orofacial dysfunction	−0.133[b]	0.004*
Age and sleep disordered breathing	−0.136[c]	0.003*

*Denotes statistically significant at $p < 0.05$.
[a] Spearman's correlation test ($r_s = 0.47$).
[b] Spearman's correlation test ($r_s = -0.136$).
[c] Spearman's correlation test ($r_s = -0.133$).

Annexure I



Nordic Orofacial Test - Screening

NOT-S

NOT-S was developed by Merete Bakke, Copenhagen; Birgitta Bergendal, Jönköping; Anita McAllister, Linköping; Lotta Sjögreen, Göteborg; and Pamela Åsten, Oslo; with the support of the Nordic Association for Disability and Oral Health, NFH.

This assessment form can be downloaded from www.mun-h-center.se.
To be used with the illustrated manual that can be ordered via Mun-H-Center's web shop or phone +46 31 750 92 00.

Nordic Orofacial Test NOT-S – screening (from 3 years)

NOT-S is used when a patient has difficulties to speak, chew, or swallow.
The anamnestic section is conducted as a structured interview. The examiner asks a question, explains, and asks additional questions when necessary, interprets the reply, and fills in the form.

The NOT-S interview contains six sections: sensory function, breathing, habits, chewing and swallowing, drooling, and dryness of the mouth (I–VI).

NOT-S examination contains six sections: face at rest, nose breathing, facial expression, masticatory muscle and jaw function, oral motor function, and speech (1–6).

The illustrated manual is to be used during the examination.

Country DK IS NO SE SF Other
Examiners Speech therapist Dentist Physician Speech therapist Other
Date of examination y y y y m m d d
Date of birth y y y y m m d d ♀ ♂
Name/ID
Primary medical diagnosis (specify only one)
Diagnostic code (ICD-10):
Examination position: seated, lying down
Position of the head when seated: normal (upright and straight), other
Answers with the help of another person

Code for screening	X = yes 0 = no – = not assessed	If there are one or more X answers in a section, place a score of 1 in the box furthest to the right.
The NOT-S total score can vary from 0 to 12		

NOT-S Total score

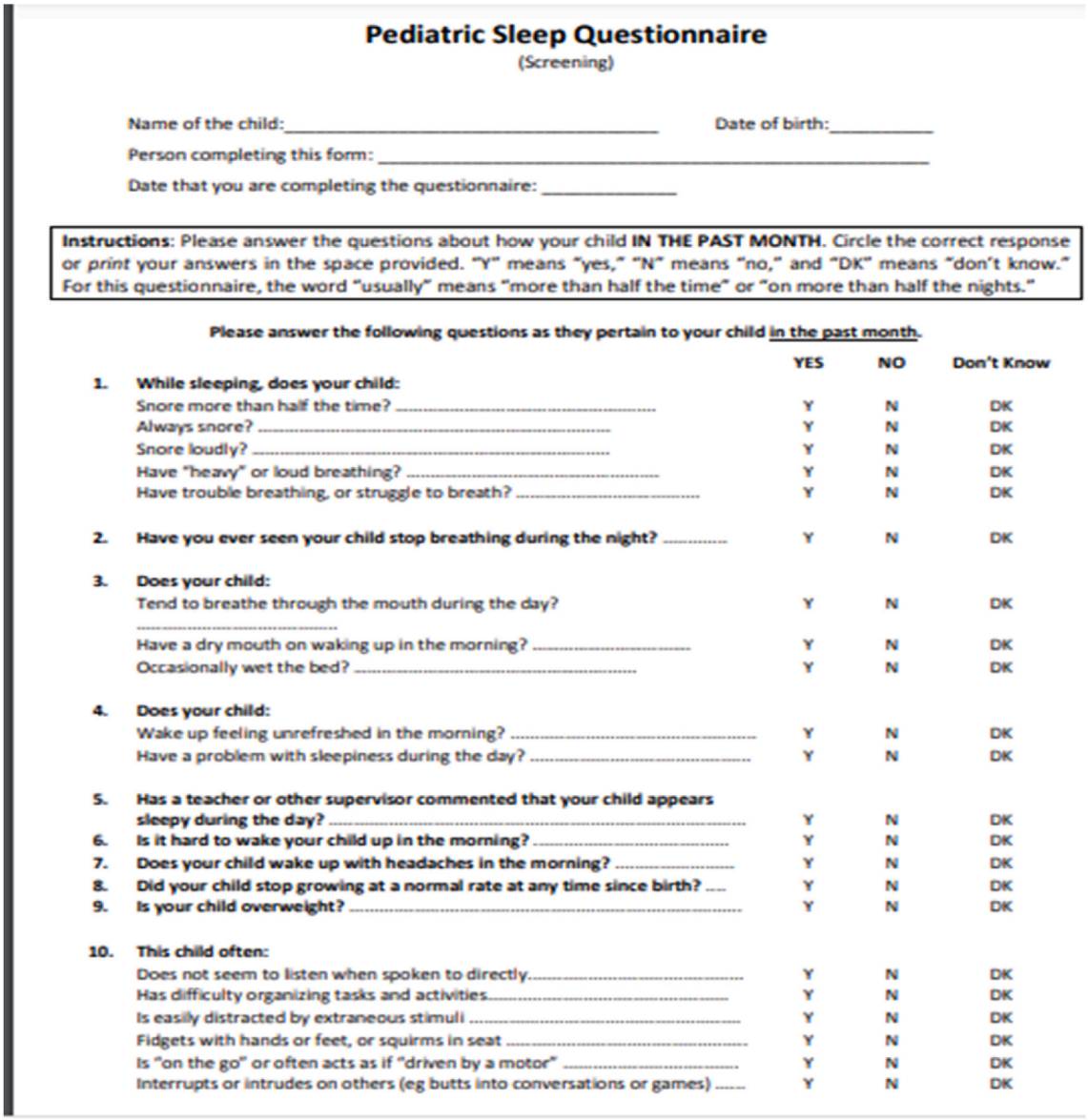
NOT-S interview

			Score
I	Sensory function		
	A. Does brushing your teeth elicit a gag reflex? Does this happen almost every time?	☐	
	Description: Obvious discomfort such as queasiness, vomiting, or refusal (increased sensitivity).		
	B. Do you put so much food in your mouth that it becomes difficult to chew? Does this happen every day?	☐	
	Description: Doesn't know when the mouth is full (decreased sensitivity).		☐
II	Breathing		
	A. Do you use any breathing support?	☐	
	Description: CPAP, respirator, oxygen, other.		
	B. Do you snore much when you sleep? Does this happen almost every night?	☐	
	Description: Snoring or apnoea. Does not apply to symptoms from asthma or allergies.		☐
III	Habits		
	A. Do you bite your nails, or suck your fingers, or other objects every day?	☐	
	Description: Use of a pacifier and sucking on the fingers is not assessed under 5 years of age.		
	B. Do you suck or bite your lips, your tongue, or your cheeks every day?	☐	
	C. Do you bite your teeth together hard or grind your teeth during the day?	☐	☐
IV	Chewing and swallowing		
	A. Does not eat with the mouth (nasogastric tube, gastrostomy or other). Skip question B–E.	☐	
	B. Do you find it difficult to eat foods with certain consistencies?	☐	
	Description: Exclude allergies and special diets such as vegetarian, vegan, and gluten-free.		
	C. Does it take you 30 minutes or more to eat a main meal?	☐	
	D. Do you swallow large bites without chewing?	☐	
	E. Do you often cough during meals?	☐	
	Description: It happens at almost every meal.		☐
V	Drooling		
	A. Do you get saliva in the corner of your mouth or on your chin almost every day?	☐	
	Description: Needs to wipe their mouth. Does not apply during sleep.		☐
VI	Dryness of the mouth		
	A. Do you have to drink to be able to eat a cracker?	☐	
	B. Do you suffer from pain in the mucous membranes in your mouth or on your tongue?	☐	
	Description: Recurrent pain or burning sensation at least once a week. Does not apply to toothache or vesicles (blister-like lesions) in the mouth.		☐
Name/ID:	NOT-S interview	Sum	☐

NOT-S examination

			Score
1	Face at rest	Watch the picture for one minute. Starting now. Observation for a total of 1 minute. Assess A-D.	
	Picture 1 A. Asymmetry	☐	
	Description: Concerns both the skeleton and soft tissues.		
	B. Deviant lip position	☐	
	Description: Open mouth or other deviations more than 2/3 of the time.		
	C. Deviant tongue position	☐	
	Description: Tip of the tongue visible between the teeth more than 2/3 of the time.		
	D. Involuntary movements	☐	
	Description: Repeated involuntary movements in the face.		☐
2	Nose breathing		
	Picture 2 A. Close your mouth and take 5 deep breaths through your nose (smell)	☐	
	Criterion: Is unable to take 5 breaths in succession through the nose. If the patient cannot close their lips, the patient or the examiner can manually help the lips to close. Do not assess if the patient has a cold.		☐
3	Facial expression		
	Picture 3 A. Close your eyes tightly	☐	
	Criterion: The facial muscles are not activated in a strongly symmetrical fashion.		
	Picture 4 B. Show your teeth	☐	
	Criterion: The lip and facial muscles are not symmetrically activated so that the teeth are easily visible.		
	Picture 5 C. Try to whistle (blow)	☐	
	Criterion: Cannot pout and round the lips symmetrically.		☐
4	Masticatory muscle and jaw function		
	Picture 6 A. Bite hard on your back teeth	☐	
	Criterion: No marked symmetrical activity can be registered when two fingers are held on the jaw muscles (the musculus masseter on both sides).		
	Picture 7 B. Open your mouth as wide as you can	☐	
	Criterion: Cannot open their mouth a distance corresponding to the width of the forefinger and the middle finger on the patient's left hand. If the front teeth are missing, use a three-finger width (the forefinger, and the middle and ring fingers) as a measure.		☐
5	Oral motor function		
	Picture 8 A. Stick out your tongue as far as you can	☐	
	Criterion: Cannot reach outside of the Vermillion border of the lips with the tip of the tongue.		
	Picture 9 B. Lick your lips	☐	
	Criterion: Cannot use the tip of the tongue to wet the lips and cannot reach the corners of the mouth.		
	Picture 10 C. "Blow up" your cheeks and hold for at least 3 seconds	☐	
	Criterion: Cannot "blow up" the cheeks without air leaking out or without making sounds.		
	Picture 11 D. Open your mouth wide and say ah-ah-ah [a]!	☐	
	Criterion: No marked elevation of the uvula and the soft palate can be observed.		☐
6	Speech A. Does not speak. Skip task B-C.	☐	
	Picture 12 B. Count out loud to ten	☐	
	Criterion: Speech is unclear with one or more indistinct sounds or abnormal nasality. Under 5 years of age, exclude R, S, and TH sounds from the assessment.		
	Picture 13 C. Say pataka, pataka, pataka	☐	
	Criterion: Do not assess this in children under 5 years of age.		☐
Name/ID:	NOT-S examination	Sum	☐

Fig. 1. NOT-S Screening and Examination tool



Pediatric Sleep Questionnaire

(Screening)

Name of the child:_____ Date of birth:_____

Person completing this form: _____

Date that you are completing the questionnaire: _____

Instructions: Please answer the questions about how your child **IN THE PAST MONTH**. Circle the correct response or *print* your answers in the space provided. "Y" means "yes," "N" means "no," and "DK" means "don't know." For this questionnaire, the word "usually" means "more than half the time" or "on more than half the nights."

Please answer the following questions as they pertain to your child in the past month.

		YES	NO	Don't Know
1.	**While sleeping, does your child:**			
	Snore more than half the time?	Y	N	DK
	Always snore?	Y	N	DK
	Snore loudly?	Y	N	DK
	Have "heavy" or loud breathing?	Y	N	DK
	Have trouble breathing, or struggle to breath?	Y	N	DK
2.	**Have you ever seen your child stop breathing during the night?**	Y	N	DK
3.	**Does your child:**			
	Tend to breathe through the mouth during the day?	Y	N	DK
	Have a dry mouth on waking up in the morning?	Y	N	DK
	Occasionally wet the bed?	Y	N	DK
4.	**Does your child:**			
	Wake up feeling unrefreshed in the morning?	Y	N	DK
	Have a problem with sleepiness during the day?	Y	N	DK
5.	**Has a teacher or other supervisor commented that your child appears sleepy during the day?**	Y	N	DK
6.	**Is it hard to wake your child up in the morning?**	Y	N	DK
7.	**Does your child wake up with headaches in the morning?**	Y	N	DK
8.	**Did your child stop growing at a normal rate at any time since birth?**	Y	N	DK
9.	**Is your child overweight?**	Y	N	DK
10.	**This child often:**			
	Does not seem to listen when spoken to directly	Y	N	DK
	Has difficulty organizing tasks and activities	Y	N	DK
	Is easily distracted by extraneous stimuli	Y	N	DK
	Fidgets with hands or feet, or squirms in seat	Y	N	DK
	Is "on the go" or often acts as if "driven by a motor"	Y	N	DK
	Interrupts or intrudes on others (eg butts into conversations or games)	Y	N	DK

Fig. 2. Pediatric Sleep Questionaire tool

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