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Development of Modified Questionnaire for Screening Purposes for Obstructive Sleep Apnea in Pediatric Population

Nikolova M[1], Raynov A[2], Rankov Kr.[1], Vicheva D[3]

[1] Department of Physiology, National Sports Academy, Sofia, Bulgaria

[2] Department of Otorhinolaryngology, University Hospital"Lozenetz", Sofia, Bulgaria

[3]Department of Otorhinolaryngology, Medical University, Plovdiv, Bulgaria

Abstract: ***Objective: To assess the relationship between the clinical complaints in pediatric population, suspected with obstructive sleep apnea (OSA), and the gravity of polysomnographic criteria, a modified protocol for screening purposes has been developed. Material and Methods: For a period of one year, during regular otorhinolaryngologic exam, five pediatric patients, aged between 4,5 and 9 years with potential obstructive sleep disordered breathing has been identified. Subsequently, their parents fill out the modified questionnaire to evaluate the necessity of further polysomnographic examination. Results: All five patients underwent split-night polysomnography and have been diagnosed with OSA, one with REM-dependent OSA. Conclusion: Based on our clinical experience, we believe that the developed protocol for screening purposes is a valuable and effective tool for early detection of pediatric OSA, while the exact diagnosis requires an overnight polysomnography as a golden standard.***

Keywords: pediatric OSA, questionnaire, screening

1. Introduction

Making a literature review on the subject the average percentage of pediatric population affected by OSA varies between 1 and 6%. It has been registered predominantly in the preschool years (3-5 years), when the hypertrophy of lymphatic tissue is more demonstrative [1] So far there is no statistically significant reported difference between both sexes, but there is a slight predominance of boys over girls up to 2-3 %[2]. As a part of the grater spectrum of Sleep Disordered Breathing (SDB) in childhood, the Pediatric OSA requires a proper screening and diagnosis. A big variety of questionnaires has been developed: Pediatric Sleep Questionnaire (PSQ) and the Child Behavior Checklist [3], Strength and Difficulties Questionnaire (SDQ), Children's Depression Inventory (CDI) and Screen for Child Anxiety Related Emotional Disorders (SCARED) [4], I'M SLEEPY [5], The Tucson Children's Assessment of Sleep Apnea Study (TuCASA) [6], etc. All this questionnaires possessed different level of complexity, reflecting cultural and language specificity making them difficult to unify and apply in the daily routine clinical practice. Our goal was to develop a simplified protocol, which reliably mirroring the gravity of the suspected OSA symptoms to the results of the subsequent polysomnographic study, used as a golden standard for diagnosis of sleep apnea.

2. Material & Methods

For a period of one year in sleep laboratory were examined five children aged from 4.5 to 9 years with suspected sleep breathing disorders. They were referred for sleep study by ENT specialist. Initially children underwent standard ENT exam, when patient with signs of acute infection of the upper respiratory tract and libratory confirmed elevated blood levels of leucocytes, C - reactive protein (CRP) and erythrocyte sedimentation rate (ESR) were excluded from the survey, to eliminate infection-induced hypertrophy of lymphatic tissue. Objectively the tonsillar hypertrophy was assessed using 4 grade scale proposed by Friedmann [7], while adenoid hypertrophy was confirmed by lateral neck radiography using Cohen and Konak method [8]. Patients with demonstrative adenotonsillar hypertrophy (ATH) were subsequently referred to PSG lab for further diagnosis, where parents completed a standardized questionnaire for respiratory disorders during sleep (E-Sleep, Ltd. questionnaire). Afterwards, all children underwent full standard overnight PSG by means of Miniscreen Pro® equipment (Heinen + Löwenstein, Germany). The PSG recordings consisted of electroencephalogram (EEG), electrooculogram, electromyogram, electrocardiogram, chest wall movements, and nasal/oral airflow and oxygen saturation. The following parameters were estimated:

- **Apnea/hypopnea index** (AHI) – number of apneas (the nasal/oral airflow cessation of at least 10 sec.) and hypopnea (airflow reduction to 50%), combined with oxygen desaturation per hour of sleep. Norm - AHI ≤ 5;
- **Desaturation index** (DI) – number of oxygen saturation drops with minimum 4% for at least 10 seconds per hour of sleep. Norm - DI ≤ 5;
- **Mean Desaturation** (Mean Des.) - mean levels of oxygen saturation drops;
- **Arousal index** (ArI) – number of desynchronizations in EEG for at least 3 sec. after previous 10 sec. sleep stage. Normal - ArI ≤ 5;
- **Sleep efficiency** (SE) - number of minutes of sleep, divided by the number of minutes in bed. Normal is approximately 85 to 90%.

The severity of OSA was determined by AHI index, as follows:

- Norm - AHI<5;
- Mild OSA - 5≤AHI<15;
- Moderate OSA - 15≤AHI<30;
- Severe OSA - AHI≥30.

These criteria are usually applied to adult patients. In children, there are still no strict criteria for OSA diagnosis because pediatric patients are few in number and they rarely get to sleep laboratory. But it is believed that one apnea per hour of sleep is sufficient for children and would lead to a decrease in oxygen saturation, which at this age can be fatal. Therefore, we will stick to these criteria (less than 1 apnea/h); nevertheless there are no officially established diagnostic criteria for pediatric OSA.

3. Results

Our primary goal was to develop a questionnaire for clinical evaluation of pediatric OSA, which reflects the early changes in sleep related symptoms throughout the night as well as child daily behavior subjectively assessed by parents. It was based on our extensive clinical experience with the scoring of Berlin and Epworth questionnaires for adult patients, and in the same time taking into consideration the specificity of kid's behavior and sleep physiology. Our questionnaire consists of 17 questions, distributed in 2 categories. The first category is focusing on the sleep disordered breathing and related symptoms. The other one is related to children daily behavior changes. Category 1 is positive if the total score equals 2 or above. The same is applicable for Category 2 - 2 or more points. The questionnaire is positive if the total score is 4 or above (23, 5%). The percentage of positive responses of these questions that show the prevalence of relevant sleep disorder was evaluated. Answers are presented in Table. 1:

Table 1: The percentage of positive answers

Children's initials	Percent of positive answers
VD	23
MW	35
TV	35
BY	40
MS	11

The presence of subjective positive indications, evaluated by the questionnaire, increases the prevalence of suspected OSA in pediatric population. Almost all children targeted by an ENT specialist for PSG study revealed presence of sleep disordered breathing.

The averaged parameters evaluated by the overnight PSG which were demonstrative for diagnosing the patients with OSA are presented in Table. 2:

Table 2: The averaged PSG parameters and standard deviation for all five children

Parameter	Average	Standard deviation
AHI (Apnea-Hypopnea Index)	22,46	8,64
DI (Desaturation Index)	19,74	1,56
Mean Des. (Mean Desaturation)	89,60	2,06
ArI (Arousal Index)	16,44	3,37
SE (Sleep Efficiency)	73,76	17,34

In all children, the registered AHI as a main diagnostic criterion was more than one per hour of sleep. Four of the children had more than 15/h, which even to the criteria for adults, meets moderate degree of OSA. All AHI were presented at Fig. 1.

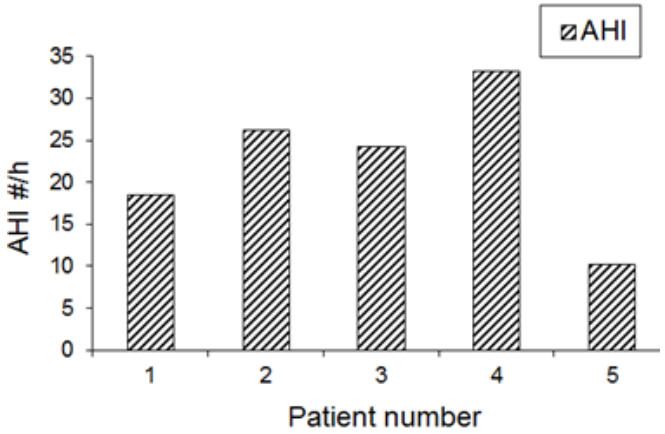

Figure 1: *AHI, #/h for children (n=5).*

One of the patients had Rapid Eye Movement (REM) sleep related OSA with AHI 18.5/h for total sleep time (TST) and 58/h for REM sleep stage (Fig. 2).

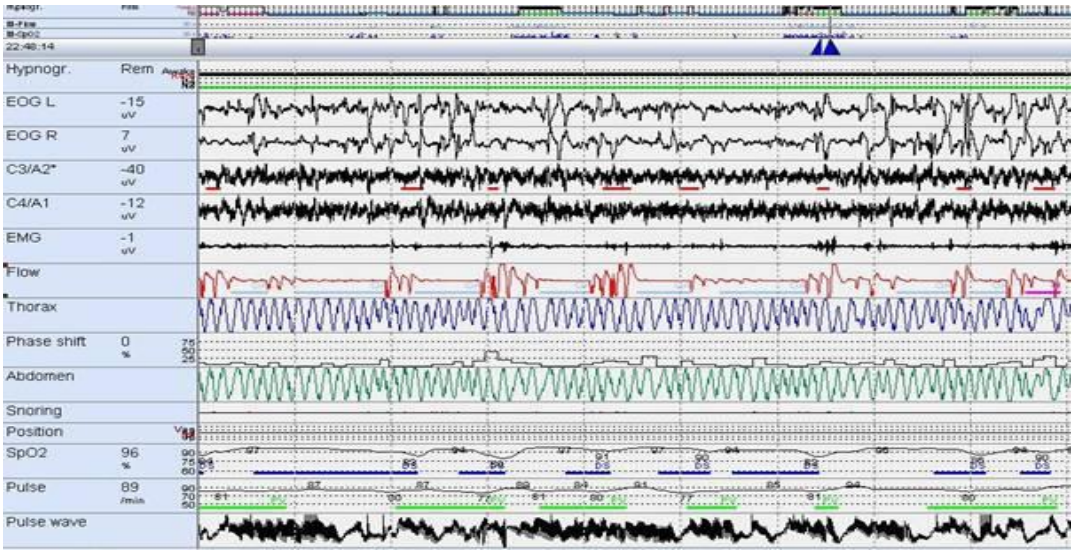

Figure 2: Row PSG data of patient with REM sleep related OSA.

The registered average oxygen saturation during wakefulness was 96.60% ± 1.02. The averaged minimal oxygen saturation (desaturation) was 89.60% ± 2.06. So, for all pediatric patients we found DI more than 5 again. This is considered as diagnostic value for adult patients. We should stick to the more strict criteria for number of oxygen saturation drops with minimum 4% for at least 10 seconds per hour of sleep. Thus DI for children should be less than 1/h same as AHI. The averaged DI for all five children was 19.74 ± 1.56.

Such an interpretation is applicable for ArI as well (ArI ≤ 1/h). For diagnosed children with OSA the average ArI was as follows: 16.44 ± 3.37.

The registered sleep efficiency for children was below normal 85% (SE = 73.76 ± 17.34). The established standard deviation was high, which means that the SE is not obligatory disturbed in all patients. Pearson correlation analysis shows low correlation coefficient (r= -0.4) between AHI and SE in children. This is probably due to the early stage of detection of sleep related breathing problems. Considering the effective screening and early detection of pediatric OSA, as well as specificity and quality of sleep in children, this result is easy understandable. To establish the reliability of the developed screening questionnaire we decided to compare the AHI, as a major diagnostic criterion for OSA, and the percentage of positive responses from the survey (Table. 3).

Table 3: The percentage of positive questionnaire's answers and AHI (#/h)

Children's initials	Percent of positive answers	AHI
VD	23	18.5
MW	35	26.2
TV	35	24.2
BY	40	33.2
MS	11	10.2

We conducted a correlation analysis between AHI and the percentage of positive responses of the questionnaire. Pearson correlation analysis shows high correlation coefficient (r= 0.95) between percent of positive answers of the questionnaire and AHI (Fig. 3).

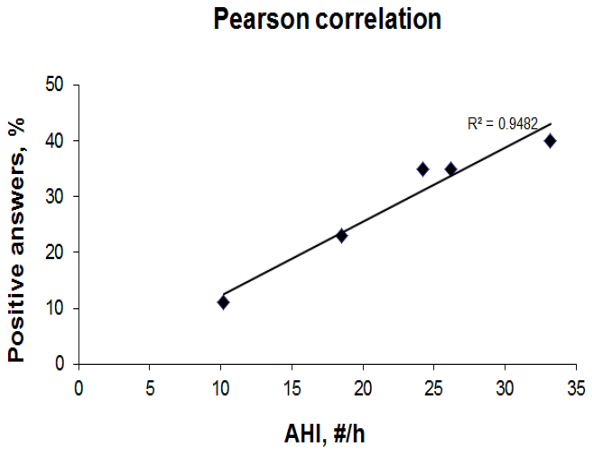

Figure 3: The Pearson correlation analysis between the percentage of positive answers of the questionnaire and AHI.

This gives us the ground to conclude that the developed questionnaire could serve as a potentially reliable screening method for early detection of pediatric OSA.

4. Discussion

There is a great variety of questionnaires for evaluation and preliminary screening of diverse sleep associated breathing disorders – Epworth [9], Berlin [10], Richland, etc. They have been developed to suit different purposes in the daily clinical work. Our goal was to build up a simplified, yet effective, questionnaire targeted both for parents and ENT specialists, who have to be the first in line medical specialist facing the problem related to pediatric OSA. The great extent of correlation between the gravity of registered sleep apnea cases and fulfillment of questionnaire criteria clearly shows the advantages of specially designed for pediatric OSA questionnaire. It's a well-known fact that Epworth questionnaire is predominantly focused on the excessive daytime sleepiness, specific for adult sleep apnea patients. On the other hand, the Berlin questionnaire is targeting only on sleep-related breathing disorders with their features in elder patients. Having a category focusing on daily behavior of children in the developed questionnaire, turns it into a unique screening tool for pediatric OSA. It is well known fact that unlike the adult patients with OSA, the children not complain of fatigue and drowsiness during the day. On the contrary, their daily behavior is manifested by attention deficit, hyperactivity, which could be considered as bad manners. This typical behavior for children with OSA makes the standard sleep breathing disorders questionnaires inapplicable in childhood. Using our combined pediatric questionnaire the clinicians will find it as a practical tool for early screening of children with suspected OSA symptoms, with regards to their specificity. Having a great extent of correlation between the gravity of sleep apnea and the positive questionnaire criteria give us ground to conclude that our initial attempts to create a standard and useful screening tool of pediatric population for sleep apnea is in the rightful direction. The timely referral of children with OSA to specialized sleep laboratories as part of a unified diagnostic-therapeutic algorithm established nationwide would be a huge contribution to adequate treatment of this disease. It is really necessary for the suspected children with OSA to be referred in time to a specialized sleep laboratory. The neglect of early symptoms of OSA leads to many unnecessary treatments and misdiagnosis. The children with OSA symptoms exhibit low levels of oxygen blood saturation which could be fatal and leads to: delays in mental and physical development, hypertension, and diabetes.

Conducting home studies for OSA, through the so-called polygraphy (PG) is not effective for pediatric population. In children sensors shift easier, which leads to artifacts and errors in the interpretation of the results. The simultaneous registration of PSG and video-recording of patient behavior through the night is possible only in the specialized sleep laboratory and this way preventing a possible equipment failure and troubleshooting, which usually accompanies home performed PG. Necessity of specialized equipment (child pulsoxymetric sensor, nasal cannula suitable for pediatric patients), which may be provided only for laboratory PSG equipment again tilts the scales in favor of

laboratory testing in children. To proper performance of the PSG study is essential to detect the flow limitations (small changes in airflow) which is possible only with nasal cannula instead thermistor sensor (usually provided in PG equipment). Otherwise the small reductions in airflow will remain misdiagnosed. The detection even of very small changes in airflow in children during sleep is essential. The gravity of impaired nasal breathing could be more precisely assessed by registering the flow limitations rather than evaluated apnoic episodes through the night, which requires PSG instead of PG. Ensuring flawless technical environment of the night study is essential for proper results and adequate sleep scoring and interpretation. The presence of skillful night technicians and sleep medicine consultants are the guarantee for correct diagnosis and further treatment of patients with sleep apnea.

The initial medical referee for sleep apnea should be a skillful medical practitioner with extensive knowledge of the primary symptoms of OSA and SDB. Great variety of medical specialties has been involved in diagnosis and treatment of the pediatric OSA. Usually children have been treated by the urologists for enuresis, endocrinologists are dealing with obesity and metabolic syndrome, and psychologists are involved in aggression and behavioral changes in school, while the main disease remaining frequently misdiagnosed. As a medical specialist who is intimately familiar with anatomy and physiology of the upper respiratory tract, the otolaryngologist is one of the most competent specialists for proper diagnosis and treatment of the pediatric obstructive sleep apnea. The thorough examination of the ear, nose and throat, the application of specific diagnostic methods (acustic rhinometry, rinomanometry, fibroendoscopy, etc) and extensive knowledge of the pathophisiology of OSA gives the otolaryngologists valuable tools for accurate diagnosis. Developing a simple, yet effective screening questionnaire for early detection of pediatric OSA could be applicable for more effective and timely discovering of the disease. Bearing in mind that in pediatric population ATH is the major factor for the upper respiratory tract obstruction defining the gravity of the OSA [11], it's reasonably to conclude that the ENT specialist is the major figure in surgical treatment. On the other hand, different medical conditions such as: muscular hypotonia, dentofacial abnormalities, neurological disorders, etc. [12], could attribute to development of pediatric OSA, requires diverse level of professional expertise and well orchestrated collaboration between different medical specialties in order to diminish the possible complications of the disease.

5. Conclusion

Developing a modified protocol for screening purposes in pediatric population give us powerful tool as medical practitioners to evaluate the initial clinical presentations of obstructive sleep apnea. The collected information is essential for ENT specialists to estimate the potential complications of OSA as a major co-morbidity factor in childhood. The prevention and early diagnosis of pediatric OSA are crucial for diminishing the negative disease outcome as well as more effective social adaptation of children with obstructive sleep apnea.

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