Sleep–Disordered Breathing in a sample of 495 children in Southern Italy

ABSTRACT

Aim Our objective was to investigate the occurrence of sleep-related breathing disturbances in a large cohort of school-aged children in Southern Italy, and to evaluate the association with anthropometric data and clinical findings of oropharynx and nasal airways.

Study design A two-phase cross-sectional study was conducted with children from schools in Turi, Italy.

Methods A screening phase aimed to identify symptomatic children and clinical data from a cohort of 495 children by a self-administered questionnaire, and an instrumental phase for the definition of sleep-related disorders and clinical analysis of oral status were performed. According to the answers, children were classified into 3 groups: habitual snorers, occasional snorers, and non-snorers. All habitual snoring children underwent a polysomnographic home evaluation, and those with oxygen desaturation index (ODI) > 2 were considered for nocturnal polygraphic monitoring (NPM). Children with apnoea/hypopnoea index (AHI) > 3 received a diagnosis of obstructive sleep apnoea syndrome (OSAS). Moreover, a complete oral examination was performed.

Results A total of 436 questionnaires (response rate: 88.08%) were returned and scored (202 M, 234 F; Mean ± Standard deviation: 6.2 ± 1.8); 18 children (4%) were identified as habitual snorers, 140 children (32%) were identified as occasional snorers, and 278 children (64%) were identified as non-snorers. The percentage of female children who were habitual snorers was higher than the percentage of male children (4.7% vs. 3.6%). Habitual snorers had significantly more nighttime symptoms. OSAS was diagnosed in 2 children by NPM. A statistically significant association between snoring, cross-bite, open-bite and increased over-jet was found.

Conclusion Habitual snoring and OSAS are significant problems for children and may be associated with diurnal symptoms. The presence of malocclusion increases the likelihood of sleep-related breathing disturbances.

Keywords: Children; Obstructive Sleep Apnoea Syndrome; Snoring.

Introduction

Although snoring often is regarded as a secondary phenomenon, it may indicate the presence of sleep apnoea or a less severe form of sleep-disordered breathing. Its prevalence varies between 3.2% and 12% depending prevalently on age [Corbo et al., 2001].

According to the International Classification of Sleep Disorders, obstructive sleep apnoea syndrome (OSAS) is a breathing disorder characterized by prolonged partial upper airway obstruction and/or intermittent complete obstruction (apnoeas) occurring during sleep, usually associated with a drop in oxygen blood saturation from 2% to 4% [Ohayon et al., 1997].

OSAS is also associated with different nocturnal and daytime symptoms [Schechter, 2002]. The clinical symptoms and polysomnographic characteristics of OSAS in children can be remarkably different from those in adults [Young et al., 1993; Brunetti et al., 2001]. Typical nocturnal signs of OSAS are breathing difficulties, forced oral breathing, sweating, troubled sleep, and unusual sleeping positions in an attempt to relieve the obstructed airways. In the daytime, children with OSAS have often behavioral problems including hyperactivity, irritability, loss of appetite, school and growth failure. OSAS can also produce cardiorespiratory consequences [Brunetti et al., 2001].

Sleep-disordered breathing usually exists in a continuum spectrum from snoring to severe obstructive sleep apnoea. Although clinical history is not sufficiently sensitive and specific to diagnose OSAS, the presence of snoring is indicative of upper airway obstruction [Schechter, 2002; Brunetti et al., 2001].

Epidemiologic data on snoring and OSAS in childhood are limited [Corbo et al., 2001; Gislason and Benediktsdottir, 1995; Bower and Gungor, 2000]. To the best of our knowledge, only one work reports epidemiologic data about these conditions in a large cohort of southern Italian children [Brunetti et al., 2001]. Our study aim was to study clinical and oral characteristics of children affected by sleep-disordered breathing in Southern Italy.

Materials and methods

Screening phase

Approval from the Provincial Education Office of Bari and Internal Ethics Committee was obtained prior to the study. The student population of Turi, a small town near Bari, was stratified by school and grade and, in each stratum, classes were randomly selected and students enrolled proportionally to the population of the town. Informed consent for enrollment was obtained by all parents. The selected sample comprised 495 children. Parents were asked to complete a self-administered questionnaire on children, and 436 questionnaire were returned. The questionnaire was devised according to Brouillette's guidelines [Brouillette et al., 1982] revised by Carroll [Carroll et al., 1995].

The questionnaire inquired about child medical history (tonsillitis, otitis, sinusitis, bronchitis, pulmonary...
infections), breathing type (oral, nasal or mixed), snoring and/or apnoeas (frequency and duration), presence of symptoms related to breathing disorders (troubled sleep, enuresis, thirst, sweating, daily sleepiness, poor school achievements). According to the results, children were classified into 3 groups: habitual snorers, when snoring occurred always or often (HS); occasional snorers, when it occurred sometime (OS); and non-snorers (NS). The questions were as follows: Does your child snore during sleep? Does your child experience apneic events during sleep? Is the child restless while asleep? Is the child irritable during the day? When your child sleeps, do you ever shake him to make him start breathing again? Do you watch your child while he sleeps because you are afraid about his breathing?

Before handling the questionnaires, in two separate occasions, parents were thought by clinicians how to recognize symptoms suggestive of snoring and OSAS. Moreover, the meaning of some words was fully explained to parents, and their definitions were included in brackets in the questionnaire.

All 436 children (202 M, 234 F; Mean age 6.2 ± 1.8) were also invited to the Dental Complex Unit for an oral evaluation, and 382 of them were eligible for the study. Dental parameters such as intercuspative line, canine and molar class, presence of cross-bite, overjet and overbite were collected and analyzed by the same operator.

Second phase
An instrumental analysis of the breathing disorder was performed in habitual snorers, for the diagnosis of OSAS or of primary snoring, by means of home instrumental tests with Vitalog HMS 5000 (Pocket Polygraph; Markos Italy); the examination was performed only when the child had no ongoing upper-airway infections. According to the guidelines of the American Sleep Disorders Association [ASDA.1994], the following parameters were monitored: transcutaneous oxygen saturation, heart rate (through ECG), snoring, and body position. For the evaluation of snoring, a microphone was positioned over the larynx. Patients with oxygen desaturation index (ODI) > 2 were regarded as suggestive of OSAS [Ali et al., 1993] and considered eligible for nocturnal polygraphic monitoring (NPM). The NPM was recorded in the sleep laboratory of the Paediatric Unit using a Vitalog HMS 5000. Polysomnography was performed overnight with constant supervision of the NPM by a physician. The following parameters were monitored: semi-quantitative airflow monitoring by means of a thermistor positioned over nostrils and mouth, heart rate (through ECG), abdominal and chest movements (by impedance plethysmography), oxygen saturation, body position (the entire NPM was audiotaped and videotaped using infrared lights), and snoring (microphone positioned over the neck). The following were also performed: EEG test, electrooculography, and electromyography. Obstructive apnoea was defined as absent airflow in the presence of respiratory effort for at least two respiratory cycle times accompanied by at least a 4% decrease in arterial oxygen saturation. Obstructive hypopnea was defined as a decrease of at least 50% in the amplitude of the oronasal thermistor signal, with maintained respiratory effort for at least two respiratory cycle times, accompanied by at least a 4% decrease in arterial oxygen saturation. Children with apnoea/hypopnoea index > 3 received a diagnosis of OSAS. The nadir of oxygen and the percentage of TST with arterial oxygen saturation < 90% were also calculated.

Statistics
The correlations between the presence of symptoms and the presence of snoring in the three categories of children were detected by $\chi^2$ test (Fisher's Exact Test). The significance level was chosen to be $p < 0.05$. The odds ratio was also performed to determine significant risk factors for habitual snoring and to verify the presence of a significant trend.

Results
Epidemiologic data
Four hundred thirty-six fully completed questionnaires were returned (436 of 495 questionnaires; 88.08%). The sample was composed of 202 male children (46.3%) and 234 female children (53.7%) with a mean age of 6.2 ± 1.8 (median: 7; range: 3 to 11 years).

Among these children 382 accepted to be evaluated at our Dentistry Unit. One hundred and sixty children (41.88%) were attending nursery schools, and 222 children (58.11%) were attending primary schools. According to questionnaires, 18 children (4%) were classified as habitual snorers (HS), 140 children (32%) as occasional snorers (OS), and 278 children (64%) as non-snorers (NS). The percentage of female children who were HS was higher than the percentage of male children, though not significantly (4.7% vs. 3.5%, respectively; $p = 0.08$). Interestingly, the frequency of apnoeas (as measured by events per night) was significantly higher in HS as compared to OS ($p < 0.0001$) (Table 1). Statistical analysis showed a higher prevalence of tonsillitis, sinusitis, otitis, pulmonary infections, the presence of rhinolalia, oral breathing, troubled sleep, enuresis, sweating, thirsty, and poor school achievements in habitual snorers as compared to occasional snorers or non-snorers (Table 1).

Oral data
Three hundred and eighty two patients underwent oral examination. Statistical analysis showed no significant association between snoring/OSAS and alteration of overjet, and molar/canine class. Statistically significant association was found between snoring, crossbite ($p = 0.0208$ HS vs. NS, $p = 0.0235$ HS vs. OS), open-bite ($p = 0.0064$ HS vs. NS), and increased overjet ($p = 0.00$ HS vs. OS) (Table 2).

Laboratory studies
All habitual snoring children (18 patients, 7 males and 11 females) underwent home instrumental evaluation. Seven children with an ODI > 2 were considered for NPM. According to parents, there was no change in clinical condition at the time of the NPM as compared to enrollment; moreover, no drugs were administered to children during the study period. In 2 cases, the NPM was
SLEEP-DISORDERED BREATHING IN CHILDREN

Repeated because of insufficient sleeping time. Two female children received the diagnosis of OSAS.

Tonsillitis (p=0.01 HS vs. NS), otitis (p<0.0000 HS vs. NS, p=0.0041 HS vs. OS), sinusitis (p=0.0001 HS vs. NS; p=0.0001 HS vs. OS), bronchitis (p=0.02 HS vs. NS), pulmonary infections (p=0.0048 HS vs. NS; p=0.0408 HS vs. OS), rhinolalia (p<0.0001 HS vs. NS; p=0.0002 HS vs. OS), oral breathing (p=0.06 HS vs. NS), enuresis (p<0.0001 HS vs. OS; p=0.0055 HS vs. OS), nocturnal sweating (p=0.0003 HS vs. NS; p=0.0013 HS vs. OS) were more frequent in children with OSAS or habitual snorers compared to children without sleep-related breathing disorders (Table 1).

**TABLE 1** - Symptoms in the three categories of children studied.

**TABLE 2** - Oral characteristics of the patients enrolled in this study.

Discussion

The present series represents one of the first study in a large cohort of Italian children. The prevalence of sleep breathing disorders ranges between 3.2% to 12.1% for habitual snoring, and between 1.1 to 2.9% for OSAS [Ali et al., 1993; Gislason and Benediktsdottir, 1995]. In southern Italy, Brunetti et al. investigated the prevalence of sleep-related breathing disorders, reporting an overall prevalence of 4.9% for habitual snoring and of 1.8% for OSAS in a cohort of 1207 children [Brunetti et al., 2001]. Several factors play a role in the definition of epidemiologic data of sleep-breathing disorders:

- sensitivity and specificity of the questionnaire used for the screening;
- adherence to the study;
- definition used for the diagnosis of OSAS [Carroll et al., 1995; Susami et al., 1971; Berry et al., 1984);
- night-to-night variation of symptoms [Witting et al., 1984];
- first-night effect [Agnew et al., 1966].

To minimize the possible confounding factors, we limited the instrumental evaluation for snoring and OSAS to the symptomatic children identified by a highly sensitive and specific questionnaire [Brouillette et al., 1982; Carroll et al., 1995]. Moreover, our diagnostic criteria are based on the most recent paediatric experiences [Carroll et al., 1995; Marcus et al., 1992; Nieminen et al., 2000] and, finally, we studied a population of children homogeneously distributed from 3 to 11 years of age with a mean age of 6.2 years. To further decrease the chances...
CAZZOLLA A.P. ET AL.

of error in the filing of the questionnaires, the medical staff accurately instructed families to recognize symptoms suggestive for OSAS and to fill in the questionnaires appropriately.

Our results demonstrate a slight female prevalence among habitual snorers but this difference is not statistically significant (p = 0.08). This data was in contrast with those reported in the literature, in which a strong male prevalence was reported [Brunetti et al., 2001; Gislon and Benediktsdottir, 1995], although this finding has not been confirmed by other authors [Ali et al., 1993; Corbo et al., 1989].

Interestingly, troubled sleep, enuresis, sweating, oral breathing, thirst, hyperactivity and poor school achievements in habitual snorers and OSAS patients were significantly more frequent than other symptoms. Moreover, the frequency of apnoeas were significantly higher in habitual snorers compared to occasional snorers and non snorers (p<0.001).

Our data support that OSAS is more frequent in snoring than in non-snoring children. However, the opposite is not true because OSAS affected only few snoring child.

Finally, using malocclusion classification based on oral examination, we showed that crossbite (p=0.0208 HS vs. NS, p=0.0235 HS vs. OS), open bite (p=0.0064 HS vs. n.s) and increased overjet (p=0.00 HS vs. OS) were important factors contributing to OSAS in children. In older patients, malocclusion might be also considered both the cause and the effect of disordered breathing.

In previous reports based on cephalometric analysis or models analysis, the typical maxillofacial characteristics of OSAS patients were identified to be a short, retro-positioned mandible, small anteroposterior airway diameter, large tongue or soft palate, low hyoid bone position, deep overbite, severe overjet [Riley et al., 1983; Lowe and Fleetham, 1991; Lowe, 1990; Lowe et al., 1986; Tsujiya et al., 1992; Susami et al., 1971]. In particular, a recent article showed a strong correlation between OSAS and malocclusion alterations such as retropositioned mandible with a strong posterior inclination of the mandibular plane [Marino et al., 2009]. Our data report, for the first time in a cohort epidemiological study on a child population, the association between OSAS and oromaxillo-facial characteristics such as crossbite.

Conclusion

Sleep-related breathing disorders in childhood shall be considered as a continuous spectrum of symptoms of variable severity, spanning from primary snoring to severe OSAS. Although they are usually considered rare disorders, they might result in important daytime problems for the child affected. It is necessary to sensitize the paediatricians toward the problem of sleep-related breathing disorders in order to identify children at risk and to offer a proper treatment avoiding unnecessary surgery.

References


