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# Prevalence of Obstructive Sleep Apnea Syndrome in a Cohort of 1,207 Children of Southern Italy\*

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Study objective: To determine the prevalence of sleep-related breathing disturbances in a large cohort of school-aged and preschool-aged children of Southern Italy.

Design and setting: This cross-sectional prevalence study was designed in two phases: a screening phase aimed to identify symptomatic children from a cohort of 1,207 by a self-administered questionnaire, and an instrumental phase for the definition of sleep-related disorders.

Patients and methods: One thousand two hundred seven children were screened by a self-administered questionnaire. There were 612 female children (51%) and 595 male children (mean age, 7.3 years; range, 3 to 11 years). According to answers, children were classified in three groups: nonsnorers, occasional snorers, and habitual snorers. All habitual snoring children underwent a polysomnographic home evaluation, and those with an oxygen desaturation index > 2 were considered for nocturnal polygraphic monitoring (NPM). Children with an apnea/hypopnea index > 3 received a diagnosis of obstructive sleep apnea syndrome (OSAS).

Results: A total of 895 questionnaires (74.2%) were returned and scored; 710 children (79.3%) were identified as nonsnorers, 141 children (15.8%) were identified as occasional snorers, and 44 children (4.9%) were identified as habitual snorers. The percentage of male children who were habitual snorers was higher than the percentage of female children who were habitual snorers (6.1% vs 3.7%, respectively; p < 0.09). OSAS was diagnosed in nine children by NPM.

Conclusion: The lower limit of prevalence of OSAS in childhood is 1% (95% confidence interval [CI], 0.8 to 1.2). If we add the five children who underwent adenoidectomy and/or tonsillectomy because of worsening clinical condition and the two children who were shown to have evidence of OSAS on domiciliary oximetry, then the prevalence is 1.8% (higher limit of prevalence; 95% CI, 1.6 to 2.0).

(CHEST 2001; 120:1930–1935)

**Key words:** childhood; epidemiology; nocturnal polygraphic monitoring; obstructive sleep apnea syndrome; primary snoring

 $\begin{array}{ll} \textbf{Abbreviations:} \ \ CI = confidence \ \ interval; \ \ NPM = nocturnal \ \ polygraphic \ \ monitoring; \ \ ODI = oxygen \ \ desaturation \\ index; \ OSAS = obstructive \ sleep \ apnea \ \ syndrome; \ TST = total \ sleep \ time \\ \end{array}$ 

A ccording to the International Classification of Sleep Disorders, obstructive sleep apnea syndrome (OSAS) is grouped among the dyssomnias and it is defined as "an intrinsic sleep disturbance characterized by the appearance of repeated episodes of upper airways obstruction (apneas) occurring during sleep, usually associated to a reduction in oxygen blood saturation." The clinical symptoms and polysomnographic characteristics of OSAS in childhood are remarkably different from those in adults.<sup>2,3</sup>

Typical signs of OSAS are breathing difficulties, forced oral breathing, profuse sweating, troubled sleep, and unusual sleeping positions in an attempt to relieve the obstructed airways. In the daytime, children with OSAS are often hyperactive, irritable, and refer loss of appetite; they may have growth disturbances and poor school achievement.

Sleep-disordered breathing exists in a continuum spectrum from snoring to severe obstructive sleep apnea; because nearly all children affected by OSAS snore, it is necessary to distinguish the two condi-

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tions. Primary snoring, a common finding in child-hood, is not associated with apnea, oxygen desaturation, or hyperventilation. However, snoring may precede sleep-related obstructive breathing disorders by many years; therefore, further investigations are often indicated.

Epidemiologic data of OSAS in childhood are limited<sup>4–6</sup>; to our best knowledge, the data herein reported show for the first time the prevalence of this condition in a large cohort of Italian children.

# MATERIALS AND METHODS

This study is a part of a large epidemiologic survey on the definition of sleep-related disorders in childhood in southern Italy, and it is aimed to evaluate the prevalence of OSAS. A sample of 1,207 children living in Molfetta, a small town with 65,000 inhabitants 30 km north of Bari, was screened in this study. The school population of this town consists of 4,867 children (1,530 in nursery and 3,337 in primary schools). The student population was stratified by schools and by year of course; in each stratum, classes were randomly selected and students in each class enrolled. The number of classes selected in each stratum was proportional to the size of stratum and sufficient to guarantee a sample size > 890, size that ensures an estimate of OSAS prevalence of 1.9 (previous estimates of OSAS range from 1.1% to 2.9%)<sup>5,6</sup> with a precision of 1% and a confidence level of 99%.

One thousand two-hundred seven children were screened, 612 children (51%) were female and 595 children (49%) were male (mean age, 7.3 years; range, 3 to 11 years); 379 children (31%) were in nursery schools and 828 children (69%) were in primary schools. This cross-sectional prevalence study was designed in two distinct phases: a screening phase aimed to select children with an history suggestive of sleep breathing anomalies and a second investigative phase. The study was carried out in accordance with the Provincial Education Office of Bari and the local school ethic committee; informed consent at enrolment was obtained by all parents.

## Screening Phase

A 41-item multiple-choice questionnaire was distributed to children at school. It was formulated according to the Brouillette's guidelines<sup>7,8</sup> revised by Carroll.<sup>9</sup> It inquired about family history for atopy, child medical history, snoring and/or apneas (frequency and duration), presence of symptoms that could be related to breathing disorders (troubled sleep, enuresis, thirst, sweating, oral breathing, need of afternoon rest, daily sleepiness, poor school achievements) and smoking habits in the family and during pregnancy (> 20 cigarettes a day). In the sleep section, questions were aimed to score the severity of sleep-related symptoms according to a 4-point scale. In the following statistical evaluation, scores of 3 (always) were defined as habitual snorers, scores of 2 (sometimes) as occasional snorers, and scores of 1 (seldom) or 0 (never) as nonsnorers. In particular the questions were as follows: does your child snore during sleep? Does your child happen to have an apnea during sleep? Is the child restless while asleep? Is the child irritable or sleepy 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 being afraid about his breathing?

Before handling the questionnaires, in two separate occasions,

parents were thought by clinicians to recognize symptoms suggestive for snoring and OSAS and to fill in the questionnaires appropriately. Moreover, the meaning of words such as apnea and/or restless while asleep were fully explained to parents; these words, when used in the questionnaire, were followed by a definition in brackets. Mothers were charged to answer the questions. Families that did not return the questionnaires were contacted by phone on two separate occasions (15 days apart) before classifying them as nonresponders.

#### Second Phase

Only habitual snorers entered the second phase of the study. Children were offered an instrumental definition of the disorder for the diagnosis either of OSAS or primary snoring. Instrumental tests were undertaken while children had no upper-airway infections. The device (Vitalog HMS 5000, Pocket Polygraph; Markos srl; Monza; Italy) was used for a nighttime home evaluation. According to the American Sleep Disorders Association relative to screening methodologies, 10 the following parameters were monitored: (1) transcutaneous saturation of oxygen, (2) heart rate (through ECG), (3) snoring, and (4) body position. For the evaluation of snoring, a microphone was positioned over the larynx. Recordings with an oxygen desaturation index (ODI) [ODI = the number of significant desaturations ( $\Delta > 4\%$ ) per hours of sleep] > 2 were regarded as suggestive of OSAS, 1,11 and children were then considered eligible for the nocturnal polygraphic monitoring (NPM). The NPM was recorded in the sleep laboratory of our unit. Clinica Pediatrica III is the tertiary referral center for the diagnosis and follow-up of sleep-disordered breathing in Southern Italy, covering an estimated population of 1,400,000 children with > 700 children followed up in our sleep clinic.

Polysomnography was performed overnight in a quiet, comfortable, darkened room in our department, with one parent sleeping in the same room with the child. A technician trained in pediatric NPM and one physician constantly supervised the NPM. No sedation or sleep deprivation was used to induce sleep. The device used for NPM was the Vitalog HMS 5000. The following parameters were monitored: (1) airflow monitored semiquantitatively by a thermistor positioned over nostrils and mouth, (2) ECG, (3) EEG (electrodes placed at A1, A2, C3, C4, and sleep stage determined by the monopolar derivation C3/A2), (4) electro-oculography (electrodes placed adjacent to the outer canthus of each eye: left outer canthus-A1, right outer canthus-A1), (4) electromyography (one electrode in the center of the chin), (5) abdominal and chest movements (by impedance plethysmography), (6) transcutaneous saturation of oxygen (by pulse oximeter waveform in order to detect motion artifact), (7) body position (the entire NPM was audiotaped and videotaped using infrared lights), and (8) snoring (microphone positioned over the neck). Recordings of < 6 h or less than two non-rapid eye movement and one rapid eye movement phases were excluded and repeated. Total sleep time (TST) was defined as the total of rapid eye movement and non-rapid eye movement sleep in a sleep episode equal to total sleep episode less awake time.

Obstructive apnea 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. The apnea/hypopnea index was calculated as (No. of obstructive apnea events plus No. of obstructive hypopnea events) per hour of sleep time. Children with an apnea/hypopnea

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 prevalence of symptoms in the three categories of snoring were compared by a  $\chi^2$  test (Fisher's Exact Test as appropriate) followed by Cochran Armitage trend test to verify the presence of a significant trend. The association between the presence of symptoms and the presence of OSAS was detected by Fisher's Exact Test, and the odds ratio was also calculated; a p value <0.05 was considered significant.

#### RESULTS

# Analysis of Questionnaires

Eight hundred ninety-five fully completed questionnaires were returned and scored (895 of 1,207 questionnaires; 74.2%). One hundred forty-three questionnaires (16%) were returned after one or two phone contacts by us; we believe that the 25% who did not respond represents the group of families that are not sensitive to the problem and therefore not are likely to have children suffering from OSAS. A comparison of demographic data of responders and nonresponders revealed no significant difference with respect to gender and age (p = not significant). Four hundred forty-one questionnaires (49.3%) were from male children, and 454 questionnaires (50.7%) were from female children. Mean age at enrollment was 7.3 years (range, 3 to 11 years). Two hundred fifty-four children (28.4%) were attending nursery schools, and 641 children (71.6%) were attending primary schools.

According to questionnaires, 44 children (4.9%) were classified as habitual snorers, 141 children (15.7%) as occasional snorers, and 710 children (79.3%) as nonsnorers. The percentage of male children who were habitual snorers was higher than the percentage of female children who were habitual snorers, though not significantly (6.1% vs 3.7%, respectively; p < 0.09). Interestingly, the frequency of apneas (as measured by events per night) was significantly higher in habitual snorers as compared to occasional snorers and nonsnorers (4 vs 2 vs 0, respectively; p < 0.01). Statistical analysis showed a higher prevalence of troubled sleep, sweating, oral breathing, hyperactivity, and poor school achievements in habitual snorers as compared to occasional snorers or nonsnorers. No difference was found in the incidence of daytime sleepiness, intense thirst at night, enuresis, and need of afternoon rest in the three categories studied (Table 1).

# Laboratory Studies

Of the 44 habitual snoring children eligible to continue the study, 5 children (12%) refused further

Table 1—Symptoms at Presentation in the Three Categories of Children Studied\*

Symptoms	NSO, % (n = 710)	OS, % (n = 141)	HS, % (n = 44)	p Value†
Apnea	0	0	25	< 0.01
Troubled sleep	13.11	25.00	47.7	< 0.01
Enuresis	1.43	3.62	4.5	NS
Thirst	9.52	12.50	11.36	NS
Nocturnal sweating	2.77	8.09	9.3	< 0.01
Oral breathing	42.0	58.99	81.82	< 0.01
Afternoon rest	11.21	14.60	4.55	NS
Daily sleepiness	1.17	3.70	4.65	< 0.05
Passive smoking	37.64	46.81	65.91	< 0.01
Smoking in pregnancy	2.59	6.43	15.91	< 0.01
Hyperactivity	13.20	20.90	25.00	< 0.05
Poor school achievement	16.48	32.62	34.09	< 0.01

<sup>\*</sup>NSO = nonsnorers; OS = occasional snorers; HS = habitual snorers; NS = not significant.

investigations following spontaneous disappearance of symptoms and 5 children (12%) underwent adenoidectomy and/or tonsillectomy before NPM because of worsening clinical condition related to apneic episodes. Surgery was followed by clinical remission in all.

The remaining 34 children underwent home instrumental evaluation; in 6 children, the recording was technically unsatisfactory and was then repeated. Fourteen children (41%) with an ODI > 2 were considered for NPM, but 2 children refused further investigation. Table 2 shows the nighttime home sleep monitoring of the 14 children with an ODI > 2and the polysomnographic characteristics of the 12 children who consented for the NPM. According to parents, there was no change in clinical condition at the time of the NPM as compared to enrollment; no drugs were administered to children during the study period. In three cases (25%), the NPM was repeated because of insufficient sleep. Of the 12 children, female children (n = 5; mean age, 8.3 years; range, 7 to 10 years) were older than the male children (n = 7; mean age, 6.5 years; range, 3.6 to 10years; p < 0.05). Nine patients (six male children; mean age, 8 years; range, 3.7 to 10 years) received a diagnosis of OSAS. Troubled sleep (p < 0.001), sweating (p < 0.01), oral breathing (p < 0.001), poor school achievements (p < 0.01), smoking in pregnancy (p < 0.01), and passive smoke (p < 0.05) were more frequent in children with OSAS as compared to children without OSAS (n = 874; the seven children who refused instrumental evaluation are excluded). Interestingly, troubled sleep (odds ratio, 19.68; 95% confidence interval [CI], 5.42 to 71.47), smoking in pregnancy (odds ratio, 10.84; 95% CI, 3.22 to 6.48), and poor school achievement (odds

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<sup>†</sup>Cochran-Armitage trend test or Fisher's Exact Test as appropriate.

Table 2—Nighttime Home Sleep Monitoring Characteristics of the 14 Children Found To Have an ODI > 2 of a Total of 34 Children Studied, and Polysomnographic Characteristics of the 12 Children Studied by NPM\*

			Home Evaluation				Laboratory Evaluation					
Case No.	Sex	Age	TST, min	ODI	Oxygen Nadir	No. of Desaturations, $\Delta > 4\%$	TST, min	ODI	A/I Index	Oxygen Nadir	No. of Desaturations $(\Delta > 4\%)$	TST With SaO <sub>2</sub> < 90%, h/min
1	M	10 yr	600	10.5	81	105	366	12.2	9.6	82	74	7/07
2	M	4 yr	700	9.1	85	106	525	11.4	7.0	85	97	4/22
3	M	8 yr	610	7.5	90	75	372	8.3	5.0	90	51	4/18
4	M	6 yr, 6 mo	675	7.0	87	79	553	6.1	3.8	85	57	5/4
5	M	9 yr	540	6.5	85	58	417.2	5.8	5.4	86	40	2/24
6	M	3 yr, 8 mo	660	6.0	84	66	460	5.1	4.1	85	38	5/12
7	F	10 yr	580	5.4	85	52	406.1	5.4	5.9	87	36	5/47
8	F	9 yr, 10 mo	584	4.8	87	47	381.2	4.0	4.2	85	26	4/09
9	F	7 yr	625	3.1	89	32	450	3.3	3.6	88	25	1/43
10	F	8 yr, 6 mo	684	3.0	90	35	481	3.7	2.7	87	30	0/85
11	F	8 yr, 10 mo	600	2.1	90	22	380	0.5	1.1	92	3	0/3
12	M	4 yr, 5 mo	680	2.4	95	27	400	0.2	1.6	95	1	0/1
13	M	7 yr, 5 mo	690	2.3	90	26						
14	F	7 yr	669	3.0	90	33						

<sup>\*</sup>M = male; F = female;  $Sao_2$  = arterial oxygen saturation; oxygen nadir = minimal value of arterial oxygen saturation during recording; TST with  $Sao_2 < 90\%$  = percentage of TST with  $Sao_2 < 90\%$ .

ratio, 3.09; 95% CI, 1.06 to 9.03) increase the likelihood of having OSAS of 19, 10, and 3 times, respectively (Table 3).

# Prevalence of OSAS

With the assumption that these nine children were the only cases of OSAS among the responders, the lower limit of prevalence in this cohort would have been 9 of 895, or 1.0% (95% CI, 0.8 to 1.2). If we add the five children who underwent adenoidectomy and/or tonsillectomy because of worsening clinical condition (diagnosed and operated in our department) and the two children who were shown on domiciliary oximetry to have evidence of obstructive apnea syndrome but who subsequently refused laboratory-based investigations, then the higher limit of prevalence of this disorder is 1.8% (95% CI, 1.6 to 2.0).

## DISCUSSION

The present study, to our knowledge the first in a large cohort of Italian children, shows an overall prevalence of sleep breathing disorders of 4.9% for habitual snoring and of 1.8% for OSAS. Interestingly, troubled sleep, nocturnal sweating, and oral breathing were significantly more frequent than other symptoms in patients with OSAS.

The prevalence of sleep breathing disorders varies widely, ranging from 3.2% to 12.1% for habitual snoring and from 1.1 to 2.9% for OSAS.<sup>4,5</sup> Several factors play a role in the definition of the prevalence of sleep breathing disorders: (1) sensitivity and spec-

ificity of the questionnaire used for the screening, (2) adherence to the study, (3) definition used for the diagnosis of OSAS, <sup>2,9,12,13</sup> (4) night-to-night variation of symptoms, <sup>14</sup> and (5) first-night effect. <sup>15</sup> To minimize the possible confounding factors, we have designed a study based on an instrumental evaluation limited to the symptomatic children identified by a highly sensitive and specific questionnaire. <sup>7–9</sup> Secondly, our criteria of using the apnea/hypopnea index for the definition of OSAS is based on the most recent pediatric experiences <sup>9,11,16</sup> that have shown that adult criteria applied to children may underestimate the prevalence of OSAS particularly in the

Table 3—Symptoms at Presentation in Children With and Without OSAS

Symptoms	With OSAS, % (n = 14)	Without OSAS, % (n = 874)	p Value
Snoring	57.1	22.6	< 0.01
Apnea	21.4	0.9	< 0.01
Troubled sleep	78.6	15.0	< 0.001
Enuresis	7.1	1.8	NS
Thirst	14.3	9.9	NS
Sweating	14.3	3.7	< 0.01
Oral breathing	100	45.8	< 0.001
Afternoon rest	0	11.6	NS
Daily sleepiness	0	1.8	NS
Passive smoking	28.6	19.3	< 0.05
Smoking in pregnancy	28.6	3.6	< 0.01
Hyperactivity	28.6	14.7	NS
Poor school achievement	42.9	19.5	< 0.05

<sup>\*</sup>See Table 1 for expansion of abbreviation.

pediatric age group.<sup>2</sup> Indeed, a separate analysis of our results clearly shows that using the adult criteria we miss four of nine diagnoses (44%) of OSAS. Finally, the diverse reported rate of sleep breathing disorders may depend on the age of the patients; indeed, the volumetric adenoids/rhinopharynx ratio varies, with the age being higher in young children thus causing obstruction easier than in older children.<sup>17</sup> In this respect, the population we have studied is well distributed across age and covers homogeneously children from 3 to 11 years of age. To further decrease the chances of error in the compilation of the questionnaires, the medical staff instructed families to recognize symptoms suggestive for OSAS and to fill in the questionnaires appropriately.

Our figure of prevalence of 1.8% is in line with the most recent data and suggests that OSAS should always be excluded in habitual snoring children. Indeed, 8 of 14 children with OSAS were habitual snorers as compared to only 196 of 874 children without OSAS (57.1% vs 22.6%; p < 0.01). According to Gislason and Benediktsdottir,<sup>5</sup> we found that snoring but not OSAS was more frequent (though not significantly) in male children; however, this finding has not been confirmed by other authors.<sup>4,18</sup> Furthermore, none of the children in our study had severe OSAS most likely because the patients at "the end of the spectrum" had already been operated on. Based on this observation and in keeping with Ali et al,4 we believe that the overall number of patients affected by OSAS should include children undergoing adenoidectomy.

Sleep breathing disorders in childhood shall be considered as a continuous spectrum of symptoms of variable severity, spanning from primary snoring to severe OSAS. In this view, the study of the upper airways resistance syndrome may be useful to complete the spectrum of the sleep breathing disorders because a small number of our primary snoring children may have had upper airways resistance syndrome; however, the need for endoesophageal pressure monitoring, a parameter not included in our study, limits the possibility of a precise diagnosis. 19,20

From the clinical perspective and family counseling, it would be important to identify predictors of OSAS. This issue has been addressed in previous studies where higher chance of having OSAS has been associated with snoring,  $^{21}$  troubled sleep, nocturnal sweating, oral breathing, poor school achievements, and daily sleepiness,  $^{5,22}$  while the role of enuresis is still controversial.  $^{23}$  In the present study, we have confirmed that snoring (p < 0.001), troubled sleep (p < 0.001), nocturnal sweating (p < 0.01), and oral breathing (p < 0.001) are positively correlated

with OSAS. Moreover, we have found that sleep is less disrupted in children with OSAS as compared to affected adults, and it might explain the absence of typical daily symptoms. Finally, we found that passive smoking (p < 0.05) and smoking in pregnancy (p < 0.001) play a pivotal role in augmenting the chance of developing OSAS, as suggested by Corbo et al  $^{18}$ 

Our data support that if a child does not snore he has limited chances of having OSAS; however, the opposite is not true because among snoring children only a few have OSAS. The inability of a clinical history to distinguish primary snoring from OSAS should prompt pediatricians and primary care physicians to start much earlier sleep disorder investigations for the definitive diagnosis.

#### Conclusion

Our study has shown an overall prevalence of OSAS of 1.8%. However, the lack of a consensus for the diagnosis of OSAS in children and the impossibility to use the adult guidelines in the pediatric contest<sup>2</sup> make it difficult to assess the prevalence of this disorder in childhood. Despite these methodologic and practical difficulties, it is necessary to sensitize the pediatrician toward the problem of sleep-related breathing disorders in order to identify children at risk and to offer a proper treatment avoiding unnecessary surgery.

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