

3-1988

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Recommended Citation

Wittig, Robert M.; Zorick, Frank J.; Roehrs, Timothy A.; Sicklesteel, Jeanne M.; and Roth, Thomas (1988) "Familial Childhood Sleep Apnea," *Henry Ford Hospital Medical Journal* : Vol. 36 : No. 1 , 13-15.
Available at: <https://scholarlycommons.henryford.com/hfhmedjournal/vol36/iss1/5>

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Familial Childhood Sleep Apnea

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We report four siblings who had polysomnographically documented sleep apnea. Two presented with the typical clinical picture of sleep apnea syndrome including daytime somnolence and snoring, had repetitive obstructive apneic episodes during sleep, and were effectively treated with upper airway surgery. The other two were asymptomatic and showed infrequent apneic episodes during sleep. This family illustrates the distinction between the sleep apnea syndrome and infrequent apneic episodes during sleep. The sleep apnea syndrome is associated with daytime symptomatology and requires treatment. The presence of apneic episodes during sleep in all four siblings has implications regarding the predisposing factors (eg, upper airway anatomy and central nervous system dysfunction) versus precipitating factors (eg, obesity, upper airway infection, and central nervous system depressants) in sleep apnea. (Henry Ford Hosp Med J 1988;36:13-5)

Obststructive sleep apnea syndrome is the most common sleep disorder seen in sleep disorders centers (1) and is described most often in adults and less often in children. The genetic or familial pattern of this disease has been a controversial issue.

The literature shows only one report of familial sleep apnea in which information was presented on five adult male siblings aged 30 to 41, their parents, and the daughter of one of the siblings (2). Three of the siblings and the father complained of excessive daytime sleepiness. This symptom is not specific to the sleep apnea syndrome but may occur with or be secondary to a variety of other conditions such as chronically insufficient sleep, narcolepsy, and medications. The differential diagnosis of daytime sleepiness often requires polysomnographic evaluation. In Strohl et al's report (2), only three of the five siblings received polysomnographic evaluation. Of the three, only one had more than five apneic episodes per hour of sleep, the most conservative number used to differentiate normal from abnormal apnea frequency. Thus, it is doubtful that the infrequent apneic episodes can adequately explain the patients' symptoms, and extrapolating data to explain the symptoms of family members who were not polysomnographically evaluated becomes even more difficult.

Distinguishing the presence of sleep apnea from a single symptom is more difficult in children than in adults. In adults daytime sleepiness is usually the chief symptom of sleep apnea syndrome, but in children the symptoms and signs are reported to be more variable (3-5) and include growth retardation, hyperactivity alternating with sleepiness, irritability, and (but less consistently than in adults) daytime sleepiness.

We report four siblings who had sleep-related apnea. Two presented with the typical clinical picture of the sleep apnea syndrome (loud snoring and daytime sleepiness) and were effectively treated with upper airway surgery. The other two had lower apnea frequencies and lacked sufficient daytime symptoms to justify aggressive intervention.

All four underwent serial polysomnographic evaluations that included continuous recording of eye movements (electrooculogram), submental EMG, and EEG (central and occipital), as well as measures of respiratory effort (thoracic and abdominal strain gauges) and nasal and oral airflow (thermistors). Most, but not all, recordings included ear oximetry using the Hewlett-Packard 47201-A oximeter. Sleep stages were scored in the conventional manner (6). Apneic episodes, defined as cessation of nasal and oral airflow for at least ten seconds, were classified as 1) central, if accompanied by no respiratory effort; 2) obstructive, if associated with respiratory effort; and 3) mixed, if both central and obstructive components were present.

Hypopneic episodes were also counted and defined as a reduction in apparent respiratory airflow or effort of at least 50% followed by an arousal. Sleep hypopnea may or may not be associated with a measurable drop (3% or greater) in oxyhemoglobin saturation (SaO₂).

Case Reports

Case 1

A 15-year-old boy was referred to us for evaluation of suspected sleep apnea syndrome. He complained of excessive daytime sleepiness (falling asleep in inappropriate places in school and at home) for about two years. He had snored loudly since the age of six. He developed enuresis at age 13, which was still a problem despite treatment with imipramine. He also complained of almost daily frontal headaches on awakening. Tonsillectomy was recommended at age nine but had not been performed. He also had seasonal (spring and fall) allergic rhinitis and had

Submitted for publication: October 20, 1987.

Accepted for publication: December 11, 1987.

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Table 1
Sleep Parameters

	Age (years)	TST (hours)	Non-REM Sleep Stages				Number of Apneas	Number of Hypopneas
			%1	%2	%3-4	%REM		
Case 1								
Preoperative	15	7.9	23	49	1	27	726	0
Postoperative	15	7.2	7	45	31	17	12	0
	20	7.6	7	51	26	16	2	16
Case 2								
Preoperative	9	8.1	6	59	22	13	151	32
Postoperative	9	8.1	5	49	28	18	5	11
	14	6.1	11	46	28	15	3	99

TST = total sleep time, and REM = rapid eye movement.

received biweekly desensitization shots for two years with partial relief of allergic symptoms. He had been a chronic mouth breather since early childhood.

On physical examination, he was 165 cm tall (30th percentile) and weighed 75 kg (149% of ideal weight). His pulse was 75 beats/min and regular, and his blood pressure was 120/80 mm Hg. He favored mouth breathing. Ear, nose, and throat examination revealed hypertrophied tonsils with only a narrow midline air space. Moderate adenoidal enlargement was also seen. There were no other anatomic findings of significance, eg, tongue, uvula, and soft palate configuration were normal.

Polysomnographic study revealed 726 primarily obstructive apneic episodes in 7.9 hours of sleep (Table 1). After adenotonsillectomy, follow-up polysomnographic study six weeks after surgery showed normal, apnea-free sleep (Table 1). Repeat study five years later documented lasting remission. At the last follow-up, he was 178 cm tall and weighed 105 kg (148% of ideal).

Case 2

This 9-year-old boy, the brother of case 1, also snored loudly and intermittently, with noticeable interruptions in breathing during sleep. He too had resumed bed wetting at the age of eight after years of nocturnal continence. Like his older brother, this was refractory to imipramine at bedtime. There were no reports of diminished school performance, although his mother thought he was excessively sleepy. His sleepiness seemed milder than his older brother's and seemed to alternate with periods of excessive activity. He also had seasonal allergic rhinitis.

On physical examination, he was 136 cm tall (75th percentile) and weighed 65.5 kg (208% of ideal). His blood pressure was 128/60 mm Hg, and his pulse was 80 beats/min and regular. His tonsils were hypertrophied but to a lesser degree than his brother's. Polysomnographic study (Table 1) showed 151 mixed and obstructive apneic episodes in 8.1 hours of sleep. The minimum SaO₂ was 67%.

After adenotonsillectomy, snoring and enuresis disappeared and daytime alertness improved. Follow-up polysomnographic study six weeks after surgery (Table 1) showed that sleep-related upper airway obstruction had disappeared. Five years later, when his height had reached 166 cm (65th percentile) and weight 128 kg (176% of ideal), his sleep apnea syndrome was still in remission.

Case 3

This patient, the only female sibling, was first evaluated at age six. Her mother complained of her snoring and somnolence. She was usually continent at night. There were no complaints of daytime sleepiness or any unusual behavior problems. She was an average student. She had

a history of atopic dermatitis, multiple allergies, and frequent upper airway infections.

On physical examination, she was 127 cm tall (95th percentile), weighed 36 kg (129% of ideal), and had hypertrophied tonsils almost meeting in the midline. Her blood pressure was 105/65 mm Hg, and her pulse was 92 beats/min.

Polysomnographic study (Table 2) showed 33 apneic episodes (66% of central origin) and 93 hypopneic episodes in 8.1 hours of sleep. No significant cardiac arrhythmias were observed. Oximetry was not available. Since there were no complaints of either disturbed sleep or daytime symptoms that might be attributable to the irregular respiration noted during sleep, no treatment was thought necessary and she was followed clinically.

When she was reevaluated eight months later, she was 131 cm tall (90th percentile) and weighed 41 kg (137% of ideal). Her tonsils, essentially unchanged, were still hypertrophied. No new symptoms were evident. She was studied in the laboratory with oximetry (Table 2). In 6.1 hours of sleep, she had 25 apneic episodes (64% mixed or obstructive) and 118 hypopneic episodes. The minimum SaO₂ was 70%. Of the hypopneic episodes, 89% were associated with more than a 3% drop in SaO₂. She was followed clinically and restudied at age nine. At that time she was 141 cm tall (90th percentile) and weighed 52.5 kg (140% of ideal). No symptoms were attributed to the irregular nocturnal respiration, and she was followed clinically.

Case 4

The youngest sibling, this 5-year-old boy was studied after his three siblings. The family said he snored but not as loudly as his siblings. His enuresis was primary, that is, he had not yet had a prolonged period of nocturnal continence. There were no complaints of daytime sleepiness or of any behavioral problems.

He was 111 cm tall (60th percentile) and weighed 23.3 kg (120% of ideal). His pulse was 90 beats/min, and his blood pressure was 110/60 mm Hg. Examination of the nose and throat was unremarkable. The tonsils were not hypertrophic, and the adenoids were small.

The results of his polysomnographic evaluation are shown in Table 2. In 7.6 hours of sleep, he had 28 apneic episodes, 65% mixed or obstructive. No cardiac arrhythmias were observed, and no treatment was thought necessary.

Three years later, he was 130 cm tall (75th percentile), weighed 31.4 kg (112% of ideal), and follow-up polysomnography showed 101 apneic episodes (98% central origin) and 171 hypopneic episodes in 9.8 hours of sleep. He was found to be suffering from acute upper respiratory infection and had taken a triprolidine-pseudoephedrine preparation (Actifed®). He was followed clinically.

Table 2
Sleep Parameters

	Age (years)	TST (hours)	Non-REM Sleep Stages				Number of Apneas	Number of Hypopneas
			% 1	% 2	% 3-4	% REM		
Case 3	6	8.1	8	44	37	11	33	93
	7	6.1	14	36	39	11	25	118
	9	5.7	25	47	24	4	18	95
Case 4	5	7.6	5	45	38	12	28	0
	8	9.8	30	42	16	12	101	171
	10	7.7	3	40	43	14	7	7

TST = total sleep time, and REM = rapid eye movement.

When he was restudied two years later, his sleep and his breathing in sleep were normal. He was 144 cm tall (85th percentile) and weighed 44 kg (110% of ideal).

The parents were queried about symptoms of sleep apnea syndrome. Both are obese, neither complains of daytime sleepiness, and only the mother was said to snore. Neither was willing to undergo polysomnographic study, but the mother was willing to make a home audio cassette recording of her snoring, which revealed loud but regular snoring without audible interruptions that might have suggested apneic episodes.

Discussion

A distinction must be made between apneic episodes during sleep and the sleep apnea syndrome. The syndrome refers to a cluster of symptoms and signs. Repetitive obstructive apneic episodes are observed on polysomnographic recording, and interventions that eliminate these frequent episodes during sleep also resolve the symptoms, as was seen in the two older siblings described in this report. When the apneic episodes are infrequent, they can be highly variable night to night (7). Apneic episodes of low frequency may not be clinically significant if they are not associated with significant hypoxemia, cardiac arrhythmia, or discernable daytime symptoms.

These siblings demonstrate a familial incidence of sleep-related apnea. Several observations can be made that may help the sleep apnea syndrome be better understood.

The two most obese siblings had the worst disease. The least overweight child (case 4) had the least apneic episodes. Case 3, with a degree of obesity in between that of her older and younger brothers, had mild apnea that has been stable over three years of growth. This is consistent with the adult experience, which sug-

gests a correlation between obesity and exacerbation of obstructive sleep apnea. The younger siblings may develop more severe apnea if they gain more weight.

Case 3 developed more frequent apneic episodes with an acute upper airway infection treated with triprolidine-pseudoephedrine (Actifed®). The apnea resolved when the illness remitted and medication was discontinued. Whether the acute airway compromise, its drug therapy, or an interaction between the two precipitated the increased apnea in sleep is unclear. Sleep apnea syndrome probably results from a combination of predisposing factors, some of which may be familial and include both genetic (upper airway anatomy, allergies, etc) and behavioral (eg, eating habits). When the condition is diagnosed, inquiry should be made about other family members who may be similarly affected.

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