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which could be correlated with its congenital origin. The etiology of Dieulafoy's disease is still unknown, and no triggering causes have been identified. However, chronic irritation, mechanical factors, and organic etiologic agents may play a role. We postulate a hypothesis based on the embryological development of the pulmonary and bronchial vessels. If the anomalous vessel originated from the pulmonary artery, this anomaly would probably have its origin from a vestige of, or abnormal development of, the distal part of the sixth aortic arch, which normally involutes on the 50th day of life. On the other hand, the vessel could proceed from an abnormal origin of the bronchial artery. This correlates with the fact that in most people the left bronchial artery derives from the aorta, unlike the right bronchial artery, which also can derive from the upper aortic intercostal and/or internal mammary arteries.<sup>6</sup> If the right bronchial artery is the source, this different development of an anomalous vessel from the aorta could be the reason for the malformation. A definite diagnosis is made by a histopathologic examination, and bronchoscopy can assess the degree of bronchial involvement. Surgery is the only cure. We agree that this malformation could be underdiagnosed; therefore, planned resections of vascular lesions could prevent biopsy procedures and complications.<sup>5</sup> The association with neurilemoma was fortuitous.

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# Attention Deficit Hyperactivity Disorder in Adults and Obstructive Sleep Apnea\*

Sohail Naseem, MD; Bashir Chaudhary, MD, FCCP; and Nancy Collop, MD

Attention deficit hyperactivity disorder (ADHD) is a common childhood illness with a prevalence between 3% and 16%. It is characterized by hyperactivity, impulsiveness, impairment in academic, social, and occupational functioning, short attention span, and onset of symptoms before age 7 years. In some patients, this illness can persist into adulthood. Obstructive sleep apnea (OSA) is a common disorder affecting 3% of the population and is characterized by hypersomnolence, snoring, disturbed sleep, and cognitive dysfunction. Some studies have suggested an association between OSA and ADHD in children. Because of the overlap of symptoms, patients with OSA may be considered to have ADHD. Significant improvement in symptoms has been reported with the use of continuous positive airway pressure in patients with OSA. We present reports of three adults who were being treated for ADHD and were found to have OSA.

(CHEST 2001; 119:294-296)

**Key words:** attention deficit hyperactivity disorder; continuous positive airway pressure therapy; obstructive sleep apnea; polysomnography

**Abbreviations:** ADHD = attention deficit hyperactivity disorder; CPAP = continuous positive airway pressure; OSA = obstructive sleep apnea

A ttention deficit hyperactivity disorder (ADHD) is a common childhood illness with a reported prevalence between 3% and 16%.<sup>1</sup> Diagnostic criteria as stated in the *Diagnostic and Statistical Manual of Mental Disorders*, fourth edition, include the onset of symptoms before age 7 years, the presence of hyperactivity, impulsiveness, short attention span, and impaired social and occupational functioning.<sup>2</sup> Sleep-disordered breathing presenting as ADHD has been suggested in several studies in the pediatric literature.<sup>3–5</sup>

Obstructive sleep apnea (OSA) is a common syndrome with a prevalence of about 3% in the general population<sup>6</sup> and 0.7 to 2.9% in children.<sup>7</sup> In adults, characteristic features are loud snoring, cognitive dysfunction, and excessive daytime somnolence. In children, the symptoms are not clear cut and can present as failure to thrive and hyperactivity, along with cognitive and learning disabilities.<sup>7</sup>

We present case reports of three adults with ADHD who were referred to a sleep center to rule out possible OSA.

## CASE REPORTS

#### Case 1

A 19-year-old white man was referred to the sleep clinic for excessive daytime somnolence and short attention span. His parents had noted loud snoring and restless sleep. Since childhood, he had experienced severe learning disabilities and dyslexia. ADHD was diagnosed in the patient, and he had been on methylphenidate therapy since age 15 years. Since being started on therapy, he had noticed improvement in his attention span with decreased daytime sleepiness.

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A physical examination revealed an obese man, weighing 105 kg. He had enlargement of the nasal turbinates, a narrowed oropharynx (Mallampati score, class III), and an enlarged uvula. The results of an examination of his cardiovascular and respiratory systems were unremarkable. His polysomnogram showed a total of 161 apneas and hypopneas with a respiratory disturbance index of 25.6 events/h. There were 162 arousals, 21 of which were due to periodic limb movements. Nocturnal oxygen saturation varied between 90% and 100%. On diagnosis of OSA, nasal continuous positive airway pressure (CPAP) therapy was initiated with significant subjective improvement in his symptoms of daytime somnolence, fatigue, and short attention span. The patient was completely weaned from methylphenidate and has not used it for > 1 year.

### Case 2

A 23-year-old white man was referred for the evaluation of sleep-related problems. The patient had received a diagnosis of ADHD a few years earlier. He had symptoms of excessive snoring, daytime somnolence, and short attention span, and occasionally he fell asleep in class. He had been receiving methylphenidate with some improvement in his symptoms.

A physical examination showed the patient to be an obese young man, weighing 105 kg. Pertinent physical findings included a thick neck with a collar size of 18 inches, a narrowed oropharynx (Mallampati score, class III). His nasal turbinates were enlarged, and his nasal septum was deviated. The patient underwent polysomnography, which showed a total of 293 apneas and hypopneas with a respiratory disturbance index of 48.5 events/h. The patient had severe nocturnal hypoxemia and spent 25% of his sleep time at < 90% oxygen saturation. He was started on nasal CPAP therapy and had significant subjective improvement in symptoms of snoring, daytime sleepiness, and short attention span. Overnight pulse oximetry showed complete resolution of nocturnal hypoxemia on 8 cm H<sub>2</sub>O CPAP. The patient was weaned from methylphenidate over the next 10 months of follow-up.

## Case 3

A 44-year-old man was referred because of very loud snoring, frequent spontaneous awakenings, fatigue and sleepiness (particularly in the afternoons), and occasional difficulty with staying awake while driving. The patient had received a diagnosis of ADHD 2 years prior to this evaluation, although he had had symptoms that were consistent with ADHD since the third grade. He was receiving treatment with a combination of dextroamphetamine and racemic amphetamine at the time of this evaluation.

A physical examination revealed an obese white man, weighing 98 kg. Nasopharyngeal examination revealed a septal deviation to the right, hypertrophied turbinates, and mild retrognathia. His Mallampati score was class II. Polysomnography showed 48 obstructive apneas and hypopneas with a respiratory disturbance index of 7 events/h. The lowest oxygen saturation was 89%. The patient elected to pursue conservative treatment including weight loss and medical therapy for nasal congestion.

#### DISCUSSION

The true incidence of ADHD in adults is unknown, but up to 2% of adults may be affected since 10 to 60% of childhood cases can persist into adulthood.<sup>8</sup> ADHD is diagnosed by the exclusion of other underlying psychiatric and neurologic disorders. Stimulant medications such as methylphenidate are prescribed for the treatment of this syndrome. Several medical conditions can present with symptoms similar to ADHD, such as thyroid disorders, petit mal epilepsy, and substance abuse. In children, sleep disorders, especially sleep apnea, have been suggested as one of the underlying causes of ADHD. In a letter to the editor, Yuen and Pelayo<sup>9</sup> stressed the possibility of underlying sleep disorders in adult as well as pediatric patients with ADHD. Although several other studies in the pediatric literature have documented this association,<sup>3–5</sup> objective data based on formal sleep studies are lacking.

Patients with OSA commonly present with loud snoring, excessive daytime somnolence, inattention, and memory difficulties. Learning disabilities and hyperactivity commonly are seen in children. Physical findings that are suggestive of this disorder are a narrow oropharynx, enlarged tonsils, swollen nasal turbinates, and morbid obesity. Unless treated, sleep disorders may have severe deleterious effects such as pulmonary and systemic hypertension, cor pulmonale, cardiac arrhythmias, and an increased incidence of cerebrovascular events.<sup>6</sup> Established treatment options include nasal CPAP, surgery, and weight reduction. Proper treatment of this disorder can effectively decrease the incidence of such complications and can improve physical and psychosocial functioning. As evident from the symptoms mentioned above, several aspects of OSA syndrome could overlap with the behavioral complex of ADHD.

To our knowledge, there are no published reports of ADHD and OSA in adults. We are reporting three such cases studied in a sleep disorders center by formal polysomnography. Our patients had life-long histories of learning difficulties along with impaired social and academic functioning. In addition, they experienced nocturnal snoring and daytime somnolence. A diagnosis of OSA was made based on the results of their polysomnography. Significant improvement in their daytime somnolence as well as psychosocial functioning was seen after therapy with nasal CPAP in two of the patients. It has been suggested that up to 25% of children who snore and who have ADHD could have their symptoms of ADHD eliminated if their sleep-related problems were corrected.<sup>10</sup> Significant improvements in symptoms after CPAP therapy and tonsillectomy in children with ADHD and underlying sleep disorders have been published by Ali and colleagues.<sup>11</sup> Once diagnosed and appropriately treated, some of their patients, as well as ours, tolerated weaning from and even discontinuation of stimulants.

There is a growing concern in the literature about the diagnosis of ADHD and escalating stimulant use. Stimulant use has been reported to have increased eightfold over the last decade.<sup>12</sup> Since methylphenidate, which is a commonly used stimulant, can improve symptoms of daytime sleepiness, its use can mask symptoms of both ADHD and OSA, thus propagating its continued use.

Based on our findings and other published studies in the pediatric literature, sleep-disordered breathing, especially OSA, should be considered in the differential diagnosis of patients with ADHD, especially those who have a suggestive history and physical findings. A correct diagnosis would significantly decrease the morbidity and mortality associated with this disorder and also would cut down on the excessive use of stimulants.

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# Aerosolized Iloprost Therapy Could Not Replace Long-term IV Epoprostenol (Prostacyclin) Administration in Severe Pulmonary Hypertension\*

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*Objectives:* To switch patients with severe pulmonary hypertension and previous life-threatening catheterrelated complications from long-term IV epoprostenol therapy to aerosolized iloprost therapy. *Design:* Open, uncontrolled trial.

Setting: Medical ICU of a university hospital. Patients: Two patients with primary pulmonary hyperpertension and one patient with pulmonary hypertension after surgical closure of atrial septal defect (mean pulmonary artery pressure  $\geq 50$  mm Hg). All were classified as New York Heart Association class II under treatment with continuous IV epoprostenol for 4 years.

Interventions: Stepwise reduction of IV epoprostenol (1 ng/kg/min steps every 3 to 10 h) during repeated inhalations of aerosolized iloprost (150 to 300  $\mu$ g/d with 6 to 18 inhalations/d). Continuous pulmonary and systemic arterial monitoring were performed.

*Results:* Aerosolized iloprost reduced pulmonary artery pressure by 49%, 49%, and 45%, respectively, and increased cardiac output by 70%, 75%, and 41% in the three patients. The effect lasted for 20 min and was similar at different doses of IV epoprostenol. Persistent treatment change to inhaled iloprost could not be achieved because all patients developed signs of right heart failure. After termination of iloprost inhalations, return to standard epoprostenol therapy led to clinical and hemodynamic restoration. *Conclusions:* Although aerosolized iloprost demonstrated short-term hemodynamic effects, it could not be utilized as alternative chronic vasodilator in patients with severe pulmonary hypertension.

### (CHEST 2001; 119:296-300)

**Key words:** aerosolized iloprost; catheter-related complications; inhaled vasodilator therapy; pulmonary hypertension

**Abbreviations:** LDH = lactate dehydrogenase; PPH = primary pulmonary hypertension

**P**rimary pulmonary hypertension (PPH) is a rare disease of unknown etiology with a progressive course, characterized by an elevated pulmonary artery pressure and pulmonary vascular resistance leading to right ventricular failure and death.1 Other forms of pulmonary hypertension may occur as a result of conditions such as collagen vascular disease, congenital systemic-to-pulmonary shunts, left-sided ventricular and valvular heart disease, respiratory disease, thromboembolic disease, portal hypertension, or HIV infection.<sup>2</sup> The responsiveness of pulmonary hypertension to vasodilators has led to the speculation that vasoconstriction is an important pathophysiologic feature of this disease. Various vasodilators have been used for longterm treatment, such as oral diazoxide<sup>3</sup> and oral calcium-channel blockers.<sup>4</sup> IV epoprostenol (prostacyclin) is one of the most potent pulmonary vasodilators, but it requires continuous IV administration due to its halflife of 2 to 3 min and, like calcium antagonists, lacks pulmonary vascular selectivity.<sup>5,6</sup> The major adverse effects of IV epoprostenol therapy are attributable to

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