

Relationship of Nasal Continuous Positive Airway Pressure to Periodic Limb Movement Disorder in a Patient Without Sleep Apnea*

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The treatment of sleep apnea with nasal continuous positive airway pressure (CPAP) occasionally has been associated with the appearance of periodic limb movement (PLM) disorder, but it has not been clear whether CPAP induces PLM, or whether the PLM is simply disguised until apnea is treated. We report the emergence of PLM after the inadvertent application of CPAP in a patient without sleep apnea.

(*Chest* 1993; 103:1612-14)

CPAP = continuous positive airway pressure; PLM = periodic limb movement

Sleep apnea and periodic limb movement (PLM) are distinctive sleep disorders but may coexist in some patients.¹ Periodic limb movement is characterized by a

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repetitive, stereotypic dorsiflexion of the toes and ankle similar to a Babinski response.² Movements typically are brief (0.5 to 2.0 s), spaced at intervals of 20 to 120 s and most prominent during non-rapid eye movement sleep. Recognition of PLM is important since it can represent a treatable cause of sleep disruption,³ but little is known about the interaction of PLM with sleep apnea and its therapy.

The treatment of sleep apnea with nasal continuous positive airway pressure (CPAP) has been associated with exacerbation of PLM disorder or the new appearance of PLM disorder in patients previously without the disorder.^{1,5} Some authors have suggested that the severe sleep fragmentation caused by sleep apnea may mask the presence of PLM, which is only revealed when apneas are controlled by CPAP.¹ Others have hypothesized that CPAP itself is a stimulus to aggravate or induce PLM.⁵ We describe a patient in whom CPAP administration induced PLM.

CASE REPORT

A 36-year-old man presented with a complaint of nonrestorative sleep and possible excessive daytime sleepiness. He had polio as a child with respiratory compromise and observed apnea, but control of his ventilation improved by young adulthood, and he could move and talk freely without effort. Nevertheless, he was reported to snore, and he weighed 225 lb at 6 ft tall. Overnight polysomnography



FIGURE 1. Normal sleep prior to CPAP. A 30-s epoch is shown. Channels 1 and 2 are EEG, channels 3 and 4 are electrooculogram, channel 5 is chin electromyogram, channel 6 is ECG, channels 7 and 8 are left and right anterior tibialis electromyogram, and channel 8 is thermistor. Three other channels are not shown.

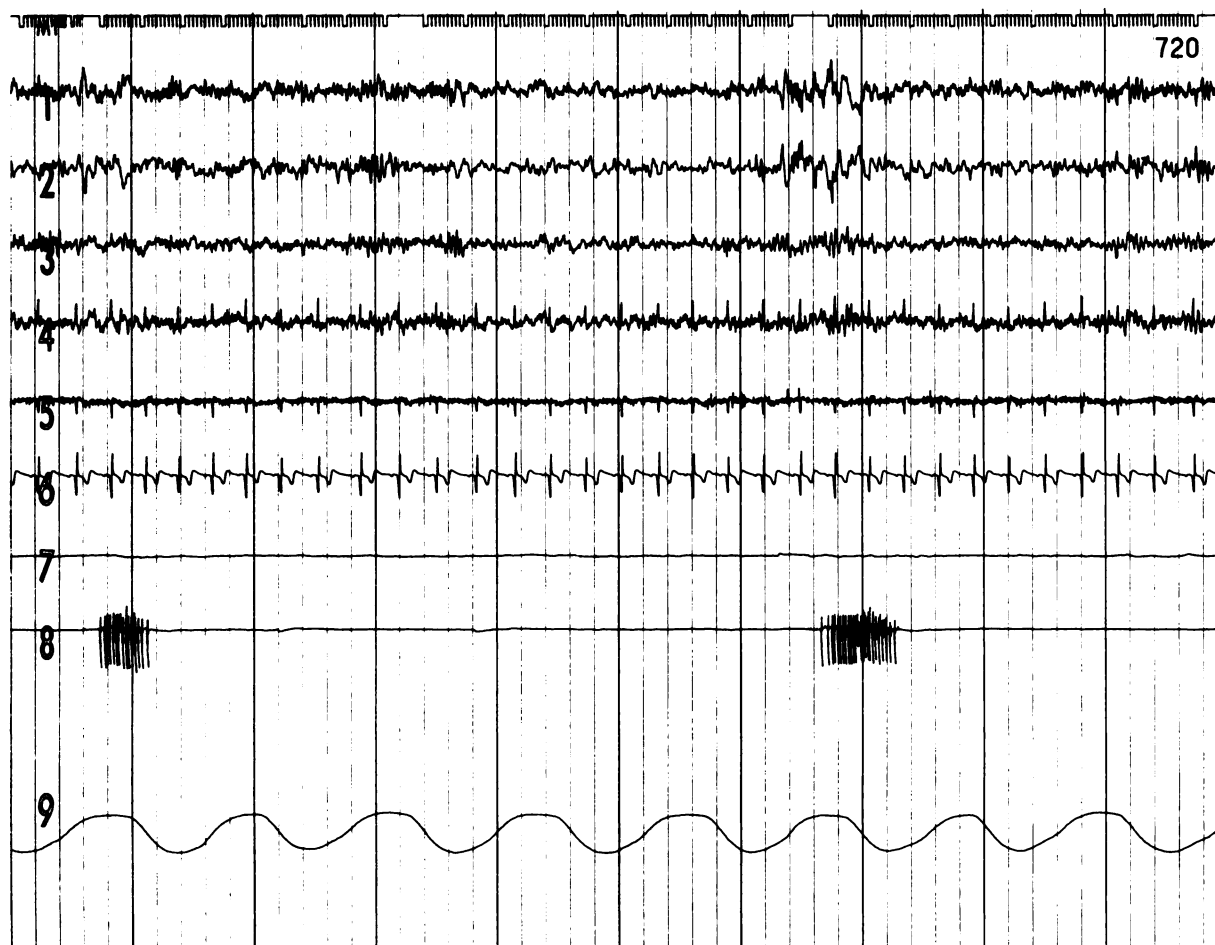


FIGURE 2. Emergence of PLM after application of CPAP. A 30-s epoch is shown. The PLM is evidenced in the right leg; PLM causes EEG arousals evidenced by a shift to faster EEG frequencies.

was scheduled for a presumptive diagnosis of sleep apnea, and CPAP was ordered for the second half of the night in anticipation of a finding of severe sleep apnea.

The polysomnogram included EEG, two-channeled electrooculogram, chin electromyogram, nasal thermistor, oral thermistor, thoracic inductive plethysmography respiratory effort, abdominal inductive plethysmography respiratory effort channels, ECG, oximetry, and bilateral anterior tibialis electromyogram for a study duration of 480 min. Resting arterial saturation was 97 percent. Sleep onset to stage 1 was observed in 38 min, stage 2 onset latency was 38 min and wakefulness after sleep onset was 91 min. Total sleep time was 351 min with a corresponding sleep efficiency of 73 percent. Sleep stage architecture was as follows: stage 1, 2 percent; stage 2, 75 percent; slow wave sleep, 1 percent; rapid eye movement, 22 percent. Reductions of airflow in both thermistors of 50 to 100 percent lasting 10 s or more were scored as apneas. Reductions in airflow of 25 to 50 percent for 10 s were scored as hypopneas.

During the first 4.5 h of the study, the patient did not have a significant degree of respiratory disturbance; his respiratory disturbance index was less than 5.0, and he was observed in all body positions. His arterial saturation was generally at 94 percent and never fell below 92 percent. There was no evidence of PLM during this part of the tracing and sleep was well consolidated with five brief arousals lasting less than 10 s (Fig 1). After 4.5 h, CPAP was applied at a pressure of 3 cm H₂O and slowly increased to 5 cm H₂O without change in any polysomnogram measure.

Further increase in CPAP pressure from 5 to 8 cm H₂O was associated with the appearance of episodes of PLM occurring regularly at 20- to 30-s intervals in the right leg (Fig 2). The episodes

of PLM were associated with brief arousals. The patient accumulated 84 episodes of PLM over 45 min (a rate of 120 per hour) before he abruptly awoke and asked that the CPAP be removed so that he could return to sleep. He then slept another 80 min with immediate resolution of the episodes of PLM after removal of the CPAP.

DISCUSSION

This patient lacked evidence of either sleep apnea or episodes of PLM prior to the application of CPAP; CPAP was associated with a high degree of PLM episodes which subsequently resolved with its discontinuation. The patient's history of polio may limit the applicability of these findings, but nevertheless these observations support the premise that CPAP *per se* can be a stimulus for PLM. In contrast, we are unaware of any reports that surgical treatment of sleep apnea is associated with PLM.

Episodes of PLM occur commonly in patients with neurologic findings or a history of neurologic disease.⁶ Although this patient had a history of polio, the absence of episodes of PLM prior to initiation of CPAP suggests that residual neurologic dysfunction due to polio was not the sole etiology of the PLM. The 20- to 40-s periodicity of PLM may be mediated by the activity of the sympathetic nervous system which also shows a 20- to 40-s periodicity.⁷ Increased activity of the sympathetic nervous system may release the PLM phenomenon.⁷ For example, psychotropic medications with an adrenergic effect have been cited as a cause of

episodes of PLM.⁸ Although the exact pathogenesis of this patient's PLM episodes is uncertain, the stress of breathing against CPAP may be a sufficient stimulus to the sympathetic nervous system to activate PLM. It is unclear whether inappropriate administration of excessive CPAP to patients with obstructive apnea induces PLM, but clinicians should recognize the potential for disrupting sleep with inappropriate CPAP administration.

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Disappearing Intracardiac Thrombi in Both Atria After Mumps in a Patient With Turner Syndrome*

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A girl with Turner syndrome was admitted with an acute cerebrovascular occlusive disease 15 days after mumps infection. Imaging techniques such as Doppler echocardiography, computed tomography and angiography of the heart revealed the existence of masses in both atria. Eight days after the last radiologic study the patient had an operation, but no masses were found in either atrium. It was thought that atrial thrombi, probably formed after viral infection, had broken down to form emboli and disappeared. It is proposed that the patients with congenital cardiopathy should be regularly examined after viral infections for possible intracardiac thrombus formation. If such a mass is found and the decision is to operate, the existence of the mass must be confirmed even in the operating room just before intervention. (*Chest* 1993; 103:1611-12)

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Since the introduction of new imaging techniques such as two-dimensional echocardiography and tomography, diagnosis of intracardiac masses is no longer problematic. In addition to neoplasms and valvular vegetations,¹ thrombi, which are often pedunculated,^{2,3} also are a cause of intracardiac masses. Because of the potential risk of pulmonary and systemic embolization^{4,5} these lesions must be surgically removed as early as possible after diagnosis. Due to the fragility of a thrombus, an intracardiac pedunculated mass may dissolve and totally disappear^{4,6} with or without signs of arterial obstruction.

CASE REPORT

A 9-year-old girl was admitted to Social Security Teaching Hospital with acute left-sided hemiplegia and right central facial paralysis. Fifteen days before the admission, she had mumps and 6 months previously she had a tonsillectomy while under general anesthesia without any bleeding problems.

She was born spontaneously after 34 weeks of gestation with a birth weight of 2,100 g. An echocardiographic study was performed at 2 years of age for the evaluation of a heart murmur and was reported to be normal. Her weight and height were below the third percentiles. Her neck was short and webbed. Cubitus valgus, widely spaced nipples and clinodactyly of the fifth finger were other findings, in addition to facial paralysis and left-sided hemiplegia. Blood pressure in the right arm was 130/90 mm Hg, but peripheral pulse and blood pressure could not be determined in the left arm and in the lower extremities. Heart auscultation revealed a grade 2 to 3 systolic murmur along the left sternal border and in the back, between the left scapula and the spinal column. The existence of the Turner syndrome phenotype, the difference in blood pressure in the extremities, and the characteristic heart murmur on the back were suggestive of coarctation of the aorta proximal to the left subclavian artery. On a Doppler echocardiogram, a pedunculated mass 18 × 18 mm in size was noted in the left atrium, and another mass was suspected in the right atrium. The left atrial mass was limiting the movement of the posterior mitral cusps but was not obstructing the mitral valve. Computed tomography (CT) supported the existence of a right atrial mass as well as a pedunculated left atrial mass (Fig 1). Right heart catheterization 22 days after hospitalization confirmed the existence of the masses in both atria. Biochemical and routine coagulation studies were within normal limits except for high levels of gonadotropins and 45 XO chromosomal pattern. Brain CT showed changes probably secondary to arterial occlusions on the right side. Eight days after catheterization,

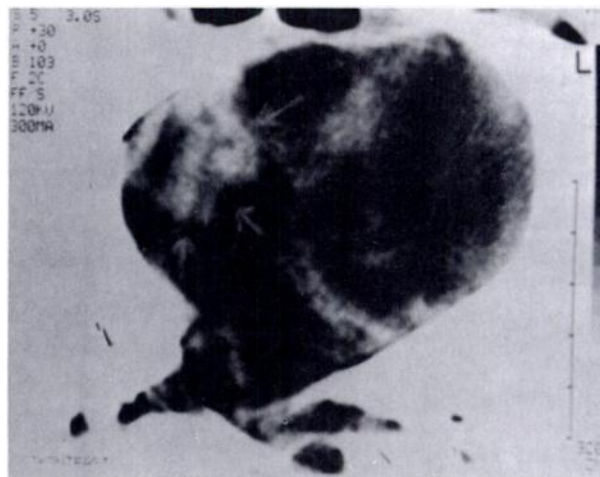


FIGURE 1. Right atrial mass and pedunculated left atrial mass (arrows).