Case Report

Uncommon cause of persistent hypoxia in a patient with obstructive sleep apnea

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Abstract

We report a case of hypothyroidism, obstructive sleep apnea (OSA) with persistent daytime hypoxemia. Cause of hypoxemia was two arteriovenous (AV) malformations in the lower lobe of the lung. We must be alert to other causes of hypoxemia in patients of OSA with persistent daytime hypoxemia.

Keywords: Hypoxemia, obstructive sleep apnoea, pulmonary AV malformations

Introduction

Congenital pulmonary AV malformation (PAVM) is a rare disease and the incidence of PAVMs is about 2.5 in 100,000.[1] Most patients are asymptomatic (60%), although signs of right to left shunt such as exertional dyspnea, cyanosis, and clubbing may be seen in severe cases.[2] So absence of symptoms does not preclude the diagnosis of PAVM.

Case Report

A 47-year-old male presented with complaints of heavy snoring and excessive daytime sleepiness for 9 years. Patient also complained of breathlessness on exertion for 6 years. He was a nonsmoker with no history of occupational exposure to dust or fumes. He was a case of hypothyroidism with bipolar mood disorder for 10 years, and was compliant with a treatment of thyroxin, sodium valproate, and resperidone.

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On physical examination, he had a BMI of 38.8 kg/m^2 and mild central cyanosis. Pulse rate was 80 beats per minute, regular and blood pressure was 120/70 mmHg in right arm, supine position.

Rest of the general physical and systemic examination did not reveal any abnormality. Pulse oximetry revealed saturation of 86–90% on room air, with higher values on supine position. ABG revealed persistent hypoxemia with hypocarbia.

Laboratory investigations revealed hemoglobin of 12.6 g%, total leukocyte count of 7100/mm^3, and a normal platelet count of 1.6 lakh. Liver function tests, lipid profile, thyroid profile, and renal function tests were all within normal limits. His chest X-ray showed no abnormality.

ECG and 2D-echocardiography were normal. Pulmonary function test revealed mild restrictive deficit. Contrast-enhanced computed tomography (CECT) of the chest suggested possibility of a pulmonary arteriovenous malformation (PAVM) in the left lower lobe [Figure 1], which was confirmed on CT angiogram [Figure 2]. An overnight polysomnography revealed severe OSA (AHI-22.5; RDI-25.4 Desaturation index-16). The patient has undergone successful embolization of the feeding

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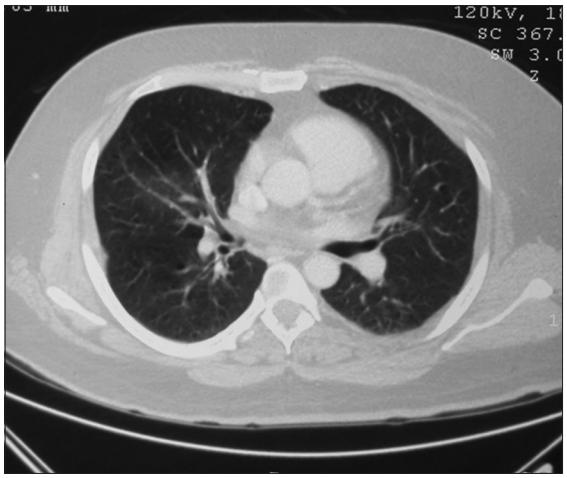

Figure 1: Contrast-enhanced computed tomography (CECT) of the chest showed pulmonary arteriovenous malformation (PAVM) in the left lower lobe

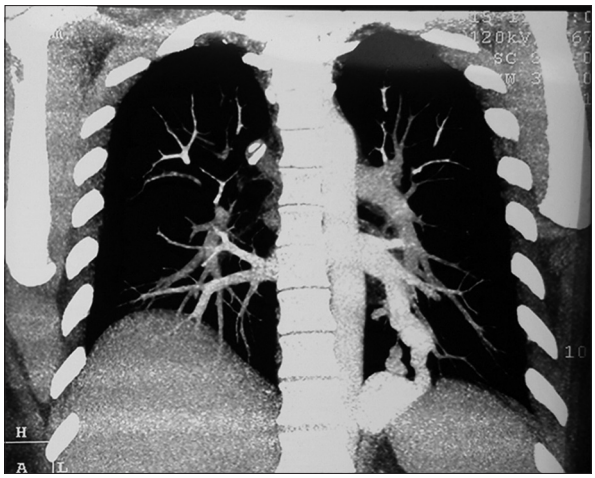

Figure 2: CT angiogram showed pulmonary arteriovenous malformation (PAVM) in the left lower lobe

artery. The patient had no clinical evidence of hereditary hemorrhagic telangiectasia (HHT).

Discussion

The association of obesity, hypothyroidism, and OSA is well established.[3] However, it is not common to have persistent daytime hypoxemia in OSA, especially in absence of symptoms. Prevalence and determinants of daytime hypoxemia in patients with OSA are not well established. However, it has been observed that patients with OSA, who spend an increased time in hypoxemia during sleep, are likely to have lower daytime PaO_2, as adjusted for age.[4] Congenital PAVM are uncommon. In one of the largest series of 15,000 consecutive autopsies, only 3 cases of PAVM were detected. However, very small PAVMs may be missed in routine autopsies. PAVMs have been reported to be twice as often in women than men.[5] Approximately 80% of PAVMs are associated with HHT.[1] Our case was a 46-year-old male who had no evidence of HHT. In the present case, there was breathlessness and cyanosis with persistent daytime hypoxemia, and the two contributing disorders were obstructive sleep apnea and congenital AV malformations. Persistent hypoxemia, even in the night time can result in pulmonary hypertension, which will not manifest in a patient with congenital PAVM, which acts as a low pressure sink.

Currently, management option of PAVM is percutaneous embolization.[6] The feeding artery may be occluded with coils. If multiple feeding arteries are present, they must all be occluded individually for successful resolution to occur.[7] Embolectomy is advisable in all PAVMs with a feeding artery diameter of over 3 mm, which should include 88% of all PAVMs, as described in Velthuis *et al.*[8] All these cases treated by embolectomy had a shunt grade of II or III.

Conclusion

Daytime hypoxemia is the hallmark clinical presentation of congenital PAVM but other disease like emphysema or interstitial lung disease should also be considered in differential diagnosis. Early differentiation from other diseases is necessary to prevent lethal complications and to institute early treatment and allow prompt recovery.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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