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Chronic migraine and transient ischemic attack due to isolated pulmonary arteriovenous malformation successfully treated with transcatheter embolization

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A 37-year-old woman, nonsmoker, with a history of chronic migraines with aura, was referred to our emergency room due to dysarthria and left faciobrachial paresis. Her family history was unremarkable. Physical examination was normal except for a mild oxygen desaturation (88% on room air). Electrocardiogram, echocardiography, peripheral venous and arterial Doppler ultrasound, and brain magnetic resonance imaging were normal. However, transcranial Doppler analysis showed a significant (shower pattern) right-to-left shunt late after isotonic saline. This pattern was present at rest and did not significantly change after Valsalva maneuver. At chest radiograph, a lobular smooth-margined opacity in the right lower lobe made us suspect the diagnosis of pulmonary arteriovenous malformation (PAVM). Chest computed tomography (CT) showed a large complex PAVM fed by two large and tortuous descending pulmonary arteries and draining through a straight vessel directed posterosuperiorly to the right lower pulmonary vein (Fig. 1). Thus, cardiac catheterization was planned to occlude the vascular malformation. The patient's informed consent to the procedure and agreement with the thoracic surgeons were obtained. The procedure was performed with local anesthesia through the femoral vein entry. Pulmonary angiography confirmed the presence of multiple large PAVMs into the right lower lobe fed by a single 1.1-cm-large vessel originating from the right inferior pulmonary artery (Fig. 2a). Transcatheter embolization of the feeding vessel was achieved with a 14-mm-large Amplatzer Vascular Plug type II (AGA Medical, Golden Valley, Minnesota, USA) device (Fig. 2b) and resulted in a sudden increase in the oxygen saturation, from 88 to 96% on room air. Clinical status and effort dyspnea significantly improved soon after the procedure and no relapse of migraine was reported. Complete occlusion of the PAVM was confirmed at follow-up CT scan (Fig. 3).

PAVM is an uncommon vascular abnormality characterized by the presence of blood vessels that allow arterial desaturated blood to shunt into the pulmonary veins without passing through the capillary bed. It may be an isolated anomaly or part of hereditary hemorrhagic telangiectasia (HTT), an autosomal dominant disorder characterized by recurrent epistaxis, mucocutaneous telangiectasias, and visceral arteriovenous malformations. Pulmonary arteriovenous fistulas of large size cause significant constrained right-to-left shunt, resulting in systemic hypoxemia and effort dyspnea. In addition, they may potentially cause paradoxical embolization [1], sharing a pathophysiologic picture similar to that of patent foramen ovalis resulting in platypnea-orthodeoxia syndrome [2–4]. Therefore, the indication for treatment of this vascular anomaly is deemed less restrictive than other potential paradoxical embolism sources [5,6]. In this setting, antiplatelet therapy is not sufficient to reduce the embolic risk due to large right-to-left shunt and, in addition, certainly does not revert any symptom caused by the constrained cyanosis. First-choice therapeutic option is nowadays transcatheter embolization with coils or other dedicated devices, which is widely deemed safer and more cost-effective than surgical lobectomy [5,6]. In patients with HTT, embolization of the PAVMs seems to decrease the prevalence of migraine attacks, according to the hypothesis that the amount of right-to-left shunt rather than its localization is involved in the pathogenesis of migraine [7]. The Amplatzer Vascular Plug type II is a new, self-expandable, cylindrical device characterized by multiple layers of occlusion that confer high occluding efficacy even in high-flow vascular malformations. Unlike other occluding devices, it can be retrieved and repositioned until a satisfactory position is achieved [8–10]. In our opinion, it could be the first-choice option in treatment of peripheral or pulmonary vascular malformations.

In conclusion, PAVM should always be ruled out as the cause of migraine in the presence of oxygen desaturation

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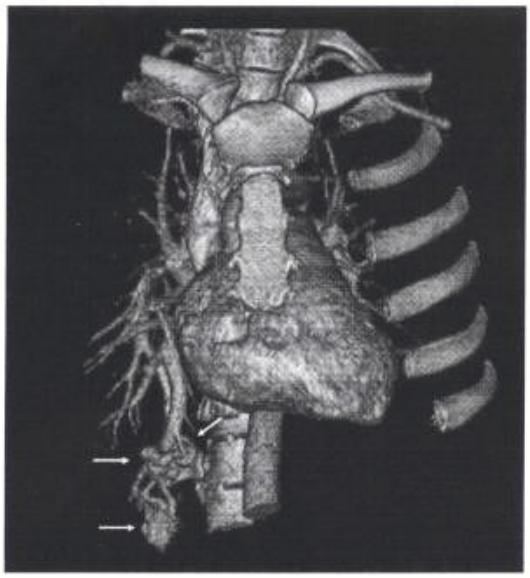
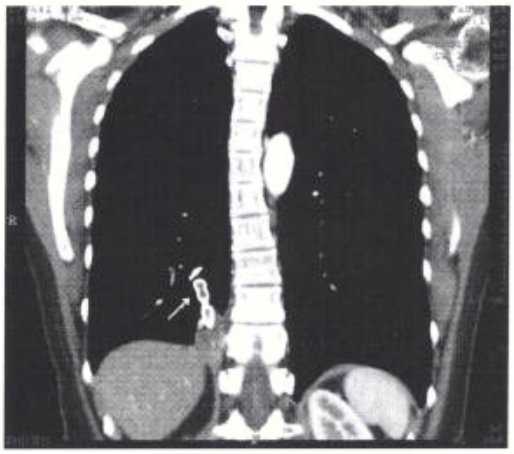
Fig. 1



Three-dimensional volume-rendering computed tomography of large right lower lobe vascular malformations (arrows) fed by large branches of the descending pulmonary artery and draining posterosuperiorly to the right lower pulmonary vein.

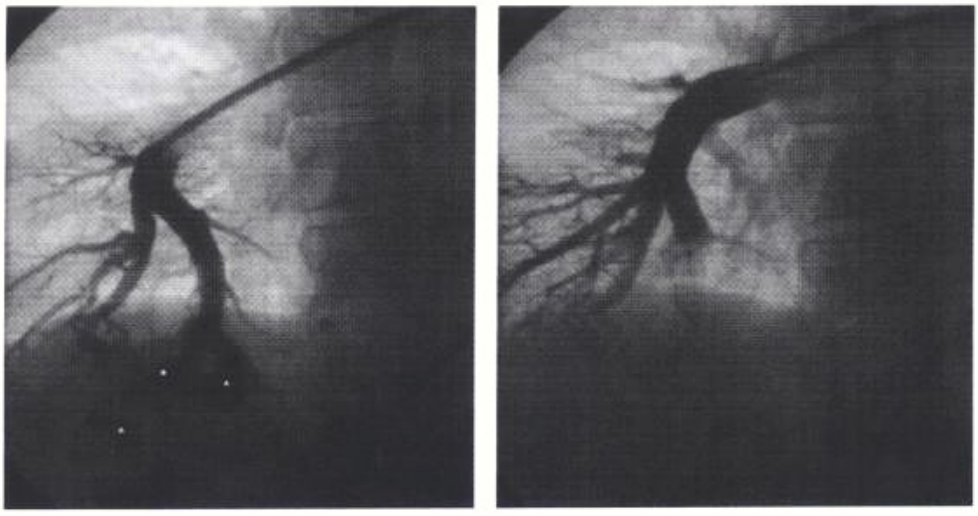
on room air, as well as a potential source of paradoxical embolization. In this malformation, percutaneous transcatheter embolization using the newest devices is safe and cost-effective with respect to surgery. However, much epidemiologic data should be collected demonstrating a favorable risk/benefit ratio of interventional treatment of PAVM before extensively supporting this therapeutic option in mildly symptomatic patients.

Fig. 3



Multiplanar reconstruction with maximum intensity projection following embolotherapy. It indicates the lack of enhancement of the pulmonary arteriovenous malformation and the presence of the occluding device inside the lower lobe (arrow).

Fig. 2



(a) Selective right pulmonary angiography shows two large feeding arteries to the multilobular vascular malformation (asterisks). (b) Total occlusion of the embolized feeding artery after device deployment.

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