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Case Report

Device closure of pulmonary arteriovenous malformation using Amplatzer vascular plug II in hereditary hemorrhagic telangiectasia



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ABSTRACT

Pulmonary arteriovenous malformations (AVM) are very rare and carry the risk of cerebral thrombo-embolism, brain abscess or pulmonary hemorrhage. The Amplatzer vascular plug II (AVP II) is a new device, used for embolization of the pulmonary AVMs. We report a case of pulmonary AVM successfully managed by using AVP II in a patient with hereditary hemorrhagic telangiectasia (HHT).

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1. Introduction

Pulmonary arteriovenous malformations are abnormal direct communications between pulmonary arteries and pulmonary veins which can be congenital or acquired. Due to high success and low complication rate, trans-catheter

closure is the preferred treatment for such malformations. A variety of devices like detachable occlusion balloon, coil embolisation and Amplatzer vascular plugs are used for embolotherapy of relatively large pulmonary AVMs. We report a case of large pulmonary AVM from left pulmonary artery (LPA) which was successfully treated with Amplatzer Vascular Plug.

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2. Case report

A 20-year old male presented with 5 years history of progressive effort dyspnea. He also had recurrent epistaxis and bleeding per rectum. There was no family history of bleeding disorders. Clinical examination showed central cyanosis, pan-digital clubbing, normal heart sounds and no murmur over the precordium. There was a short systolic murmur audible in the left infra axillary area increasing with deep inspiration and Muller's maneuver. Chest skiagram showed a homogenous opacity in the left lower zone (Fig. 1A). Electrocardiogram was normal. Trans-thoracic echocardiogram showed normal biventricular function and no obvious intracardiac structural anomalies. There was an echolucent area postero-lateral to the left ventricle (Fig. 1B). Contrast echo with agitated saline showed filling of this echolucent space followed by dense opacification of left sided chambers, a few cardiac cycles after appearance of contrast in the right heart chambers (Fig. 1C, D, E). Computerized tomography (CT) of thorax showed an AVM close to the left ventricle (Fig. 1F). In retrospect, oxygen saturation in lying down position was 90% with worsening of desaturation on standing up (82%) suggestive of platypnoea ortho-deoxia syndrome. In view of pulmonary AVM and recurrent epistaxis, hereditary hemorrhagic telangiectasia (HHT) was suspected. Further evaluation with CT scan brain and abdomen didn't reveal any AVMs, aneurysm or infarcts. Nasal endoscopy showed multiple arteriovenous malformations of nasal mucosa (Fig. 2A). Upper gastro-intestinal endoscopy and colonoscopy didn't demonstrate any telangiectasia. Pulmonary angiogram showed large AVM arising from descending branch of left pulmonary artery (Fig. 2B, C, D). No significant feeders (>3 mm) which may necessitate coil embolisation after device closure of main vessel were identified. Angiographic studies of celiac axis, superior and inferior mesenteric arteries were normal. The patient fulfilled the Curacao criteria for hereditary hemorrhagic telangiectasia (HHT) as he had spontaneous recurrent epistaxis, multiple telangiectasias in oral mucosa and proven visceral AVM of lung.²

As the patient was not keen for surgery, device closure with AVP II was planned. A 6-Fr Goodale-lubin (G L) catheter was advanced into left pulmonary artery (LPA) and 0.035" Backup Meier wire introduced through GL catheter into the AVM. A 9-Fr ASD sheath was subsequently advanced over the wire into lower branch of LPA. A 22 mm AVP II was introduced through the sheath and deployed in the lower division of LPA. Repeat angiogram didn't show filling of the AVM and SpO2 improved to 98%. The patient is asymptomatic on follow up.

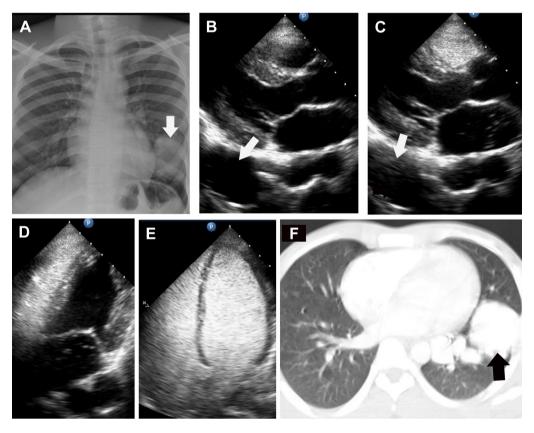


Fig. 1 – (A) X-ray chest showing homogenous opacity in right lower zone. (B) Trans-thoracic echo: parasternal long axis (PLAX) view showing the echolucent space (AVM) posterior to left ventricle. (C) Contrast echocardiogram using agitated saline in PLAX view filling the right ventricle and the AVM before reaching the left sided chambers. (D) Contrast echocardiogram in apical four chamber view shows filling of the AVM situated lateral to left ventricle. (E) Dense filling of entire left heart during contrast echocardiogram with agitated saline. (F) Contrast enhanced CT-thorax showing pulmonary AVM.

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