Successful lobectomy for central large pulmonary arteriovenous malformation

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Abstract

A pulmonary arteriovenous malformation (PAVM) is caused by abnormal communications between the pulmonary arteries and veins. In this study, a 64-year old woman presented with a large PAVM in the central upper lobe of the right lung. As feeding vessels were large and short, the patient was scheduled for resection therapy. By clamping the right main pulmonary artery, the blood flow into the PAVM was controlled and lobectomy was performed safely. Although advances in interventional radiology have led to the introduction of obliterative techniques, surgical resection is still an effective first option for patients with a large, centrally located PAVM.

Keywords: Arteriovenous malformation • Pulmonary • Thoracic surgery

INTRODUCTION

A pulmonary arteriovenous malformation (PAVM) is caused by abnormal communications between the pulmonary arteries and veins [1]. This malformation has a right-to-left shunt and induces chronic hypoxaemia, paradoxical embolism or infection. A large, centrally located PAVM with aneurysmal formation is uncommon and the optimal treatment remains unclear [2].

CASE REPORT

A 64-year old woman with a PAVM in the upper lobe of the right lung, initially discovered several decades ago, was referred to our hospital with recently developed symptoms including dyspnoea on exertion, cyanosis, polycythemia and right cardiac failure. There was no family history of hereditary haemorrhagic telangiectasia. Significant clinical findings included central cyanosis and digital clubbing. The laboratory results included a low systemic arterial oxygen saturation (SaO₂) of 65% and a haemoglobin level of 20.4 g/dl. Chest radiography revealed a lobulated mass in the upper right lung, and chest computed tomography (CT) showed a 10 × 8-cm nodular lesion centrally located in the upper lobe of the right lung (Fig. 1a). A pulmonary angiogram confirmed the lesion to be a PAVM supplied by feeding arteries 8 mm in diameter and 1.5 cm in length before entering the venous sac (Fig. 1b). The shunt fraction was calculated as 53% by a whole-body scan with technetium-99m-labelled macroaggregated albumin particles.

The patient underwent a right anterolateral thoracotomy. Findings at surgery included a dilated superior pulmonary vein (Supplementary Video 1; Fig. 1c) and a large pulsating complex

PAVM, which nearly completely occupied the right upper lobe. The PAVM was supplied by the superior trunk of the right pulmonary artery. To avoid damaging the weakened vessel walls, we clamped the right main pulmonary artery and controlled the blood flow into the PAVM (Fig. 1d). Restriction of the blood flow into the right upper lobe significantly reduced the diameters of both superior trunks and superior vein, which allowed for safe stapling (Fig. 1e and f). Furthermore, SaO₂ improved from 63 to 100% immediately after clamping the right pulmonary artery (Table 1). Pathologically, a cystic structure with an irregular lumen and anastomosing vascular channels was noted. The post-operative course was uneventful. A follow-up examination at 1-year showed no cyanosis or dyspnoea on exertion, and room air arterial blood gas oxygen saturation of 98%.

DISCUSSION

Transcatheter embolization, which involves blocking of the artery that leads to the PAVM with metallic coils or detachable balloons, is the treatment of choice and most lesions can be successfully managed [3, 4]. However, failures of transcatheter embolization for PAVMs occur more often in aneurysms with large and short feeding vessels because of a high risk of inadvertent coil migration as well as a significant instability of the guiding catheter during coil deployment [5]. Georgiou *et al.* [6] reported that, in the presence of a large solitary malformation, centrally located, surgery is still a safe and effective option. In the present case, after considering both the vessel diameter and length, interventional therapy was thought to be inappropriate as treatment.

Recently, video-assisted thoracoscopy has been used during resection of a small PAVM [4]. In the present case,

Figure 1: (a) Chest CT scan showing arteriovenous fistula (arrows). (b) Pulmonary angiography showing pulmonary arteriovenous malformation located in the right upper lobe with the short length of the feeding artery before entering the venous sac (arrow). (c) Photograph of operative field. Dilated superior pulmonary vein. (d) Main pulmonary artery and dilated superior trunk of the right pulmonary artery. (e) Dilated superior pulmonary vein before clamping of main pulmonary artery (yellow arrow). (f) Collapsed superior pulmonary vein after clamping of main pulmonary artery. Blocking blood flow to the right main pulmonary artery significantly reduced the diameter of the superior pulmonary vein (yellow arrow).

Table 1: Perioperative arterial blood gas (ABG)

	FiO ₂ (%)	pH	pCO ₂ (mmHg)	PO ₂ (mmHg)
Preoperative (baseline)	Room air	7.39	34.1	41.0
Intraoperative preresection	100	7.28	51.9	55.4
Intraoperative postresection	100	7.29	50.1	400
Postoperative (POD 7)	Room air	7.39	34.6	75.9

the large size (8 cm in diameter) and the position near the hilum implied that the blood flow into the PAVM was extremely high. In addition, some investigators have proposed that the cause is a defect in the terminal arterial loops [7], which causes enlargement of thin-walled capillary sacs, indicating that the feeding vessels as well as aneurysm sac might be fragile. Therefore, we used a muscle-sparing thoracotomy approach to achieve better hilar and vascular control. In addition, we considered that there was potential for damage to the pulmonary artery or pulmonary vein

Supplementary Video 1: A dilated superior pulmonary vein was seen at the hilar region of the right upper lobe. To avoid damaging the weakened vessel walls, we clamped the right main pulmonary artery and controlled the blood flow into the PAVM. Restriction of the blood flow into the right upper lobe significantly reduced the diameters of both superior trunks and superior vein, which allowed for safe stapling the superior vein (V1-3) and superior trunk of the right pulmonary artery.

during transection of those vessels, therefore we blocked the blood flow into the PAVM before shearing off the vessels. Using this method, a right upper lobectomy was safely performed.

In conclusion, we report a case of a large PAVM with aneurysmal formation located in the pulmonary hilum. Surgical treatment still plays an important role when managing such large, high-flow PAVMs with aneurysmal formation in a central location.

SUPPLEMENTARY MATERIAL

Supplementary material is available at *ICVTS* online.

Conflict of interest: none declared.

REFERENCES

- [1] Khurshid I, Downie GH. Pulmonary arteriovenous malformation. *Postgrad Med J* 2002;78:191–7.
- [2] Hsu PK, Hsu WH, Wu YC. Central large pulmonary arteriovenous malformation with aneurysmal dilatation. *Thorac Cardiovasc Surg* 2009;57: 434–6.
- [3] Pollak JS, Saluja S, Thabet A, Henderson KJ, Denbow N, White RI Jr. Clinical and anatomic outcomes after embolotherapy of pulmonary arteriovenous malformations. *J Vasc Interv Radiol* 2006;17:35–44.
- [4] Gupta P, Mordin C, Curtis J, Hughes JM, Shovlin CL, Jackson JE. Pulmonary arteriovenous malformations: effect of embolization on right-to-left shunt, hypoxemia, and exercise tolerance in 66 patients. *AJR Am J Roentgenol* 2002;179:347–55.
- [5] Tal MG, Saluja S, Henderson KJ, White RI Jr. Vein of Galen technique for occluding the aneurysmal sac of pulmonary arteriovenous malformations. *J Vasc Interv Radiol* 2002;13:1261–4.
- [6] Georgioudis GP, Berman M, Vidne BA, Saute M. Pulmonary arteriovenous malformation treated by lobectomy. *Eur J Cardiothorac Surg* 2003;24: 328–30.
- [7] Watanabe N, Munakata Y, Ogiwara M, Miyatake M, Nakagawa F, Hirayama J. A case of pulmonary arteriovenous malformation in a patient with brain abscess successfully treated with video-assisted thoracoscopic resection. *Chest* 1995;108:1724–7.