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Surgical Treatment of Pulmonary Arteriovenous Malformations: A Case Series

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Abstract

PAVM is a rare but fatal disease in many cases. Patients may be asymptomatic or present dyspnea, cyanosis, and polyglobulia. However, complications such as paradoxical thromboembolism, brain abscess, and massive hemoptysis are causes of higher morbidity and mortality. This article presents 3 patients with PAVM who underwent lobectomy between 2014 and 2018. The clinical and surgical data were obtained from their medical records. In the 3 cases, pulmonary lobectomy was performed, 2 received hybrid treatment: previous embolization and pulmonary surgery. The mean age was 41.3 years. The main symptom was cyanosis. In 1 case the affection was in a single lobe; in 2 cases it was complex. We consider that surgery is the first treatment option, especially when the PAVM is localized and greater than 5cm or when transcatheter embolization cannot be performed.

Keywords: Pulmonary arteriovenous malformation • Polyglobulia dyspnea • Lobectomy

Introduction

Pulmonary arteriovenous malformations (PAVMs) are direct communications between the branches of the pulmonary arteries and veins, without an intermediate lung bed [1]. The incidence in the general population is estimated at 2 to 3 cases per 100,000 inhabitants. They may be asymptomatic or present with dyspnea, cyanosis, polyglobulia, and clubbing [2]. We present 3 cases of patients with PAVM underwent open surgery with previous embolization. The data consigned in this case report were extracted from the clinical history of the patients with PAVM, who received surgical treatment at Guillermo Almenara Hospital. Likewise, the patients authorized and consented to the publication [3].

Case Series

Case 1

A 48-year-old woman with a history of hereditary hemorrhagic telangiectasia (Rendu Osler Weber's disease) and pulmonary tuberculosis. She presents peripheral cyanosis and clubbing. 1 year before surgery, she began to present hemoptysis of 50 cc in volume. A chest CT scan reveals a large pulmonary arteriovenous malformation located in the upper lobe of the right lung and 2 smaller PAVMs in the lower lobe of the left lung (Figures 1A and 1B).

Consequently, the patient underwent embolizations with coils of the PAVMs located in the left lower lobe because they were small (Figures 1C and 1D). However, the main lesion was in the right upper lobe, this lesion could not be resolved by endovascular route because the caliber of the afferent artery was greater than 10 mm, so we decided to have pulmonary surgery. Upon admission, oxygen saturation was 80% with $FiO_{2=}$0.21. She is scheduled for a right upper lobectomy for vertical axillary thoracotomy (Figures 1E and 1F).

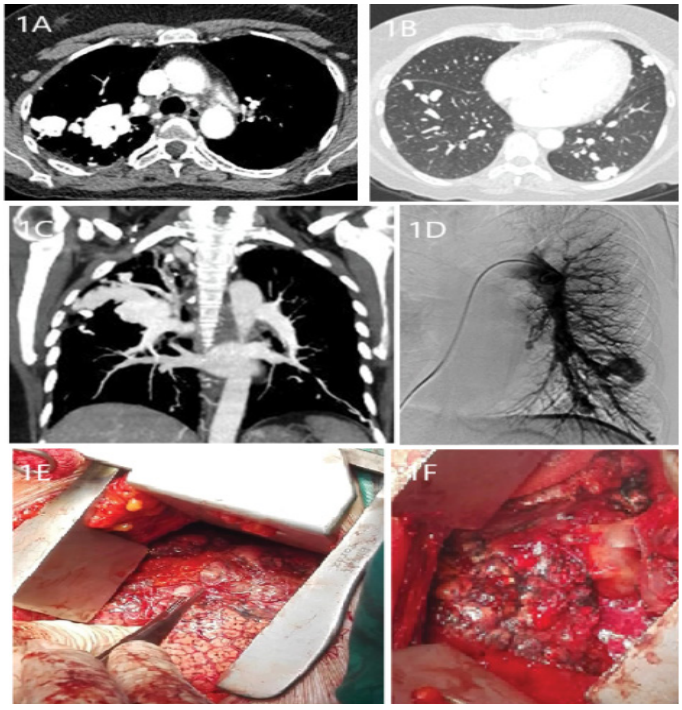

Figure 1. (A and B) Chest CT angiography: PAVM is in the upper lobe of the right lung and other smaller PAVMs in the lower left lobe. **(C and D)** Chest angiography. **(E and F)** Vertical axillary thoracotomy shows PAVM. Complex pulmonary PAVMs with aberrant vessels of the anterior trunk artery in the right upper lobe.

Pulse oximetry improved after resection of the affected lung lobe. She was discharged on the seventh day after the intervention with oxygen saturation at 96% (FiO_2=0.21).

Case 2

A 30-year-old woman with a history of hereditary hemorrhagic telangiectasia (Rendu Osler Weber's disease). She presents central cyanosis and clubbing. 1 year before the surgery, after her last pregnancy, she began to present hemoptysis and was hospitalized. The chest CT angiography shows a large PAVM located in the lower lobe of the right lung and multiple others in the left lung of a smaller size and peripheral location. Upon admission, oxygen saturation was 85% with FiO_2=0.21. In the first instance, it was decided to

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carry out embolizations of the AVMs located in the left lung because they were small: however.

Small PAVMs of the left lung were successfully embolized; however, the giant PAVMs of the lower lobe of the right lung did not have a favorable anatomy for embolization. Likewise, this AVM had as tributaries, branches of the pulmonary artery and visceral and parietal arteries from the abdominal aorta artery.

Therefore, prior to surgery, parietal artery embolization is performed: intercostal arteries, right internal mammary artery and visceral branches from the celiac trunk that fed the giant malformation (Figures 2A - 2F)

We decided to opt for surgery: right lower lobectomy. In the postoperative period, she presented a retained hemothorax and was reoperated for exploratory thoracotomy plus revision of hemostasis. She was discharged two weeks after surgery with 95% oxygen saturation with FiO_2=0.21.

Case 3

A 46-year-old woman presented, during her last pregnancy (8 years before her lung surgery) episodes of lipothymia and central cyanosis, as well as oxygen desaturation (up to 80% with FiO_2=0.21). 2 years before admission, the patient presented paresthesia in the right upper limb, associated to headache and fever, so she was hospitalized for a transient cerebral ischemic attack and brain abscess (Figures 3A and 3B).

She underwent pulmonary arteriography, this exam showed that PAVMs dependent on arterial branch of the segment 6 and venous drainage towards the vein of the right lower lobe (Figures 3C and 3D). During surgery, an increase in oxygen saturation (97%) is evident after resection of the affected lung lobe. In the postoperative period, she evolved without complications and was discharged on the sixth day after the intervention with 98% oxygen saturation with FiO_2= 0.21.

Discussion

The mean age was 41.3 and all were female. 2 out of 3 cases had a diagnosis of Rendu Osler Weber syndrome (ROW). The most frequent symptom is dyspnea followed by cyanosis. In our case series, threatening hemoptysis was presented in two cases. Also, in 1 of the 3 cases, PAVM diagnosis was made to identify the brain abscess etiological agent. Likewise, most of these 2 were complex and 1 simple. We had no cases of AVM of subpleural location [4]

CT angiography is currently the most widely accepted non-invasive method [4] Primary or congenital PAVM are the most frequent [5]. On the other hand, 2 patients presented anemia and 3 had polyglobulia. Embolization is recommended for PAVM with feeding arteries greater than 3 mm in diameter. Recent studies show that the risk of brain complications such as stroke and brain abscess is independent of the diameter of the alimentary artery [5].

In our experience, it was used successfully in complex PAVM, as a hybrid treatment. On the other hand, it should be mentioned that due to its size, AVM can be divided into small (less than 5 cm) or large (more than 5 cm). We had 3 cases larger than.

In 3 cases, improved post-intervention oximetry, and this improvement was maintained for up to 12 months. Likewise, the dyspnea also improved after surgery and embolization.

In all cases, we performed pulmonary lobectomy due to PAVM and in two of these cases embolization was performed prior to surgery, either to reduce the tributaries of the alimentary artery or to treat peripheral AVMs in the contralateral lung [5].

All three cases presented complications such as hemoptysis. However, in the third case, the patient had a stroke (paradoxical embolism) and even brain abscess (severe polyglobulia) that worsened his quality of life (Table 1). However, if patients are operated on early, that is, once the pulmonary arteriovenous malformation has been determined, the risk of complications decreases considerably. In our hospital, there is not always material and supplies to perform endovacular treatment (embolization), for this reason we believe that surgery is a current treatment option for this pathology, especially in localized cases with unfavorable anatomy for endovascular treatment.

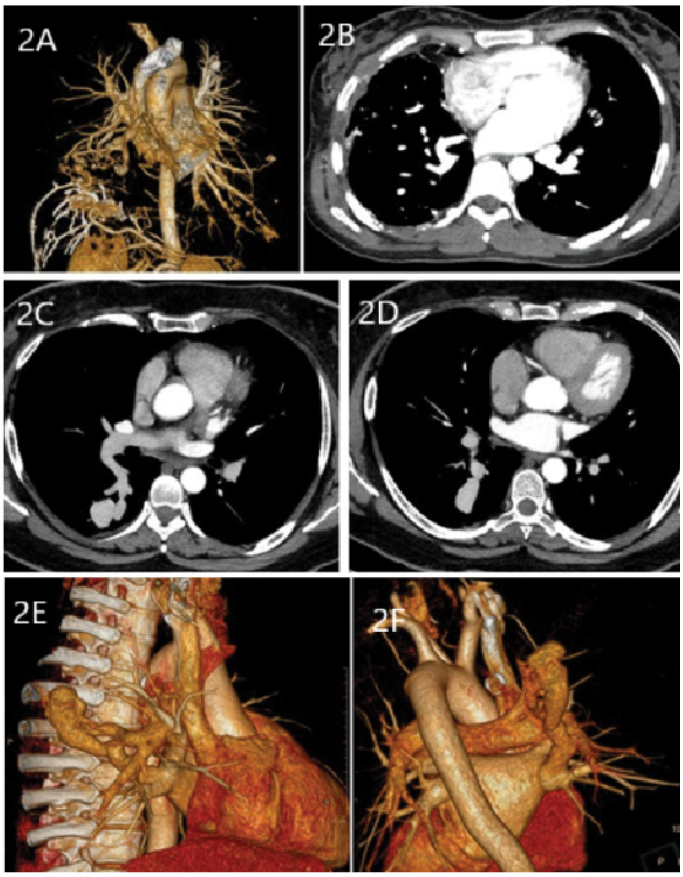

Figure 2. (A and B) Chest CT angiography: Note that the vascular clew is fed by parietal arteries (intercostal arteries) and visceral arteries (celiac arterial trunk). Image on the right shows a chest tomography on postoperative day 12. **(C and D)** Note that the vascular bundle is dependent on the pulmonary artery branch and venous drainage is directed towards the pulmonary vein in the left atrium. **(E and F)** Chest CT angiography with reconstruction.

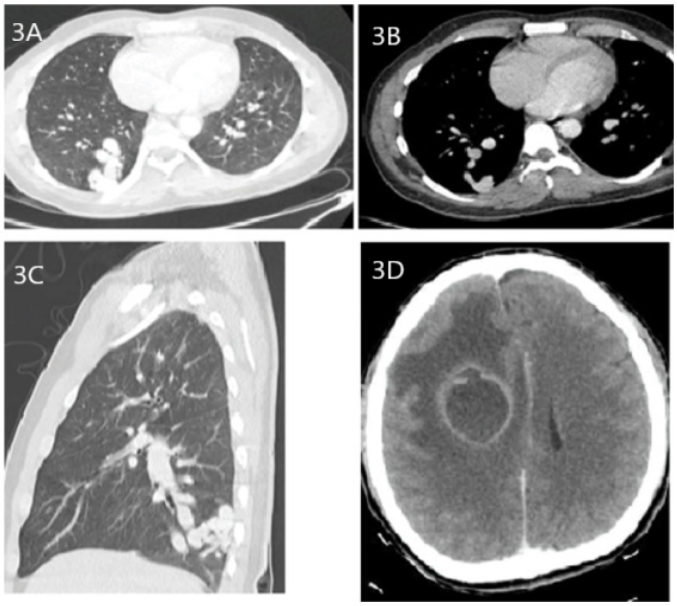

Figure 3. (A and B) Chest CT angiography: PAVM is in the lower lobe of the right lung and **(3C -3D)** Brain CT, transient cerebral ischemic attack and brain abscess is evidenced.

Table 1. Clinical characteristics of 3 patients with a diagnosis of pulmonary arteriovenous malformation.

N	Gender	Age	Main symptoms	Other AVM	History	Complications
1	F	48	Dyspnea Cyanosis Polyglobulia	Brain and pancreas	EROW Tuberculosis Hypothyroidism	Hemoptysis
2	F	30	Dyspnea Cyanosis Polyglobulia		EROW Hyperinsulinism	Hemoptysis
3	F	46	Dyspnea Cyanosis Polyglobulia		Stroke Brain abscess	Paradoxical thromboembolism

F: Female, M: Male, EROW: Rendu Osler Weber's Disease, AVM: Arterio- Venous Malformation

Conclusion

We consider that the treatment of PAVM is surgery as the first option, especially when the PAVM is localized and greater than 5 cm or when transcatheter embolization cannot be performed due to unfavorable vascular anatomy. In some patients with complex PAVM or there is some complication (paradoxical embolism and brain abscess), a hybrid treatment can be performed; that is, reduction of the vascular malformation with embolizations and later performing pulmonary lobectomy.

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