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Pulmonary venous aneurysm: Forming a glove balloon-like shape

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ABSTRACT

INTRODUCTION: Pulmonary venous aneurysm (PVA) is a candidate for surgery because of the risk of rupture and continued growth even if the patient does not develop mitral valve disorders.
PRESENTATION OF CASE: We report the case of PVA that continued to increase in size throughout a 17-year observational period.
DISCUSSION AND CONCLUSION: The location and form of the aneurysm are important to consider when choosing the surgical approach. If the aneurysm forms a "glove balloon-like shape," lobectomy is necessary because end-to-end anastomosis is difficult to perform in such cases.

1. Introduction

Pulmonary venous aneurysm (PVA) is a rare venous abnormality characterized by local aneurysmal dilation of the pulmonary vein. PVA can be of an acquired or a congenital origin. If acquired, PVA is usually associated with mitral valve disease. In most cases of PVA, patients are asymptomatic and surgical treatment is not required unless there are complications or PVA becomes enlarged. Here we describe a case of PVA, wherein the aneurysm slowly enlarged throughout a 17-year observational period. Lobectomy was required because of the shape and location of the PVA.

2. Case report

A 64-year-old female with right upper PVA diagnosed 17 years earlier presented with chest pain at rest. On admission, her vital signs, results of initial laboratory tests, and electrocardiographic and echocardiographic findings were unremarkable. There were no signs of coronary artery diseases or other internal diseases, such as mitral valve disorders. However, a chest roentgenogram revealed a mass on the hilum of the right lung and chest computed tomography (CT) indicated aneurysm growth over the past 10 years (from 45 mm to 52 mm in diameter). Three-dimensional reconstruction of the CT scan (3D-CT) showed that each segmental pulmonary vein flowed into the aneurysm independently and the aneurysm was located at the base of the right upper pulmonary vein (Fig. 1A,B). The aneurysm became enlarged; thus, surgical intervention was chosen to prevent rupture. Intraoperative findings revealed an enlarged saccular aneurysm with a very thin wall (Fig. 2). As revealed by 3D-CT, each segmental vein flowed into the wall of the aneurysm; thus, it was impossible to perform end-to-end anastomosis. The distance between the right middle lobe vein and the aneurysm was sufficient and there was no venous stricture. Venous resection was possible at the proximal side of the right upper lobe vein, which had a normal venous wall. Because the middle lobe vein could be conserved, the upper lobe was resected with the aneurysmal upper lobe vein. Pathological examination of the resected specimen revealed degeneration of the pulmonary venous wall but no clear pathogenesis (Fig. 3). The patient's postoperative course was uneventful, and she was discharged on the postoperative day 9. No recurrence was detected 2 years after surgery.

3. Disscussion and conclusion

PVA is a rare venous abnormality characterized by aneurysmal dilation of the pulmonary vein with only 75 cases reported by 2014 [1]. In our case, the aneurysm was followed up for 17 years. To the best of our knowledge, this is the longest follow-up period for a PVA.

The etiology of PVA remains uncertain. Although acquired PVA is usually associated with regurgitation of the mitral valve and chronic pulmonary hypertension, the cause of idiopathic PVA remains unclear. Our patient did not have heart disease or mitral valve disease and her chest pain was accidental. Although the aneurysm wall consisted of a normal series of layers, the elastic fiber layer had various thicknesses and partially tore or thinned.

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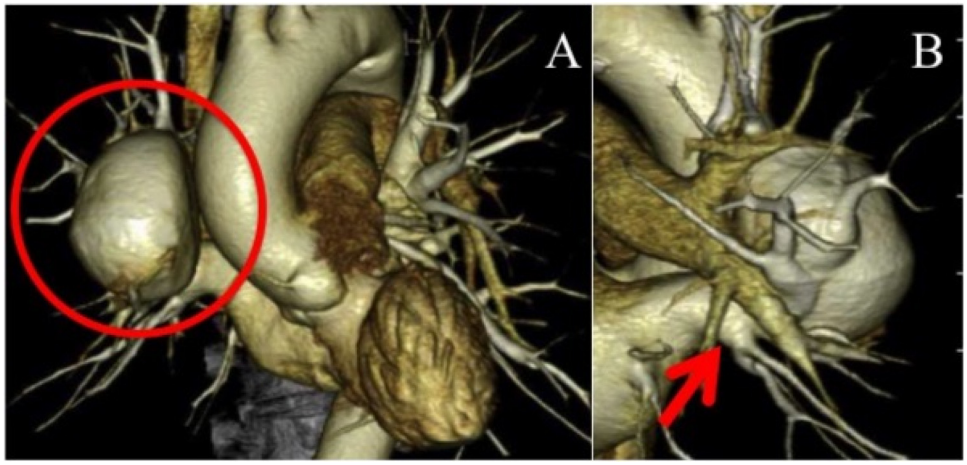

Fig. 1. A) 3AD-CT demonstrating a large pulmonary aneurysm (circle). B) EBach segmental pulmonary vein flowing into the aneurysm independently. The distance between the right middle lobe vein and the aneurysm was sufficient to be free from stricture. The right upper lobe vein was resected at normal venous wall and the middle lobe vein could be conserved (arrow).

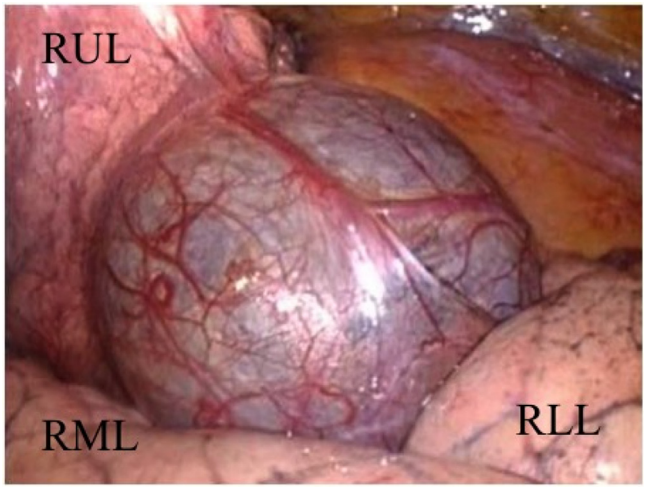

Fig. 2. Operative findings demonstrating an enlarged pulmonary venous aneurysm with very thin wall. PVA: pulmonary venous aneurysm. RUL: right upper lobe. RML: right middle lobe. RLL: right lower lobe.

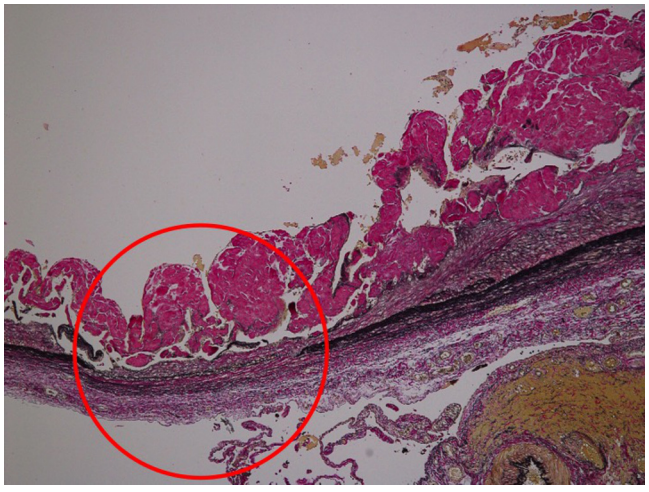

Fig. 3. Histology of pulmonary venous aneurysm wall with elastic-van Gieson stain. The elastic fiber layer tore partially (circle).

This degeneration of the layer was possibly a direct cause of the aneurysm, but then again was possibly a result of the aneurysmal change. The origin of the aneurysm was unclear; thus, her PVA was deemed idiopathic.

PVA can be classified into three morphological categories: saccular type, tortuous type, and conflict type [2]. Of the reported cases in the literature, 62% of aneurysms of the conflict type and 19% of the tortuous type were associated with valve disease, but no saccular type case was associated with valve disease. Our case was classified as saccular type; thus, an idiopathic PVA likely developed into a saccular aneurysm.

Computed tomographic angiography, echocardiography, and angiography are essential for an accurate diagnosis of PVA. In the literature, intraoperative transesophageal echocardiography was employed to demonstrate the shape of the PVA and details of pulmonary venous connections [3]. Preoperative information is very important to choose an appropriate surgical approach, and 3D-CT is the optimum modality to evaluate PVA preoperatively. Surgical treatment is necessary only in cases with complications, such as rupture of the aneurysm, which can be fatal. In the literature, size of PVA undergone surgical intervention varied. David and colleagues reported a case of PVA with a diameter of 55 mm and Alexander and colleagues reported a case of PVA with a diameter of 100mm[1,3]. At present, there is no consensus on the frequency of rupture, size criteria for surgery, or the optimal time for surgery for PVA. For congenital PVA, even if not associated with mitral valve disease, the aneurysm becomes inherently unstable and slowly increases in size, requiring thorough long-term observation after diagnosis. Careful follow-up is important and if PVA shows a tendency to enlarge, surgical intervention should be chosen to prevent rupture.

For surgical treatment, pulmonary vein continuity by end-to-end anastomosis and azygos vein patch reconstruction can be considered, but these procedures require sufficient margins at the distal and proximal stumps of the pulmonary vein for anastomosis. In our case, the aneurysm formed a “glove balloon-like shape” and was located at the base of the right upper pulmonary vein, and each normal segmental pulmonary vein flowed into the aneurysm directly, just like an inflated glove balloon. In conclusion, for a PVA with a “glove balloon-like shape,” lung resection is recommended.

Conflicts of interest

The authors declared that there is no conflict of interest.

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Ethical approval

Not requested.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contribution

Critical revision: Jun Hanaoka, Yasuhiko Ohshio, Masayuki Hashimoto

Guarantor

Kazuki Hayashi

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