## Case Report

# A Case of Pulmonary Artery Aneurysm Related to Pulmonary Valve Regurgitation

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A 56-year old woman was admitted to our institution for the treatment of pulmonary artery aneurysm and pulmonary valve regurgitation. Multislice three-dimensional computed tomography showed a very large (61 mm in diameter) pulmonary artery aneurysm. Transthoracic echocardiography revealed severe pulmonary valve regurgitation and an enlarged right ventricle. Pulmonary artery aneurysmorrhaphy and pulmonary valve replacement were performed. Postoperative hemodynamics were stable. No recurrent dilatation of the pulmonary artery or cardiac failure was observed 9 months after surgery.

#### ACTA MEDICA NAGASAKIENSIA 65: 25-28, 2021

Key words: Pulmonary aretery aneurysm, pulmonary valve regurgitation, pulmonary valve replacement, aneurysmorrhaphy

### Introduction

Pulmonary artery aneurysm (PAA) is a rare disease with unclear history and prognosis and no clear guidelines about the surgical indications are available. The surgical argument in favor of correction focuses on the prevention of pulmonary hypertension and aneurysm rupture. We report a case of a main PAA related to pulmonary valve regurgitation.

### **Case Report**

A 56-year old woman was admitted to our institution for the treatment of PAA and pulmonary valve regurgitation. She did not have cardiac failure or any other subjective symptoms. Multislice three-dimensional computed tomography showed a large (61-mm diameter) PAA (Figure 1,2). Transthoracic echocardiography revealed severe pulmonary valve regurgitation and an enlarged (53 mm) right ventricle. The mean pulmonary arterial pressure of 11 mmHg was within the normal range. Median sternotomy and standard cardiopulmonary bypass were performed. An aortic cross-clamp was applied, and antegrade cold blood cardioplegia was initiated. The main PAA was easily identified. Under cardiac arrest, the pulmonary artery was incised longitudinally up to the bifurcation of the right and left pulmonary arteries. The tricuspid pulmonary valve showed no signs of stenotic lesions, such as degeneration, induration, or fusion of the leaflets. Besides, neither perforation nor deviation was observed. However, marked annulus dilatation was noted. The pulmonary valve was replaced with a 25-mm bioprosthetic valve. A direct visual inspection found the pulmonary arterial wall to be well-preserved, with no dissection or marked thinning. At the incision site, the right and left sections of the anterior wall of the main pulmonary artery were resected, and aneurysmorrhaphy was performed to reduce its diameter to approximately 3 cm. The patient was weaned off cardiopulmonary bypass without complications. Pathological examination of the pulmonary

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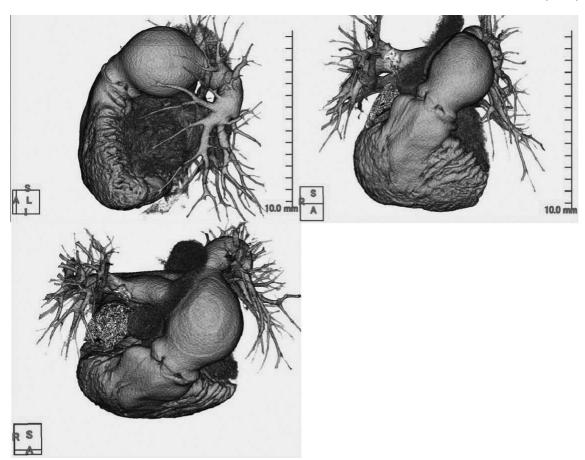


Figure 1. Three-dimensional reconstruction of chest computed tomography. Shown here is diffuse aneurysmal dilation of the pulmonary artery.

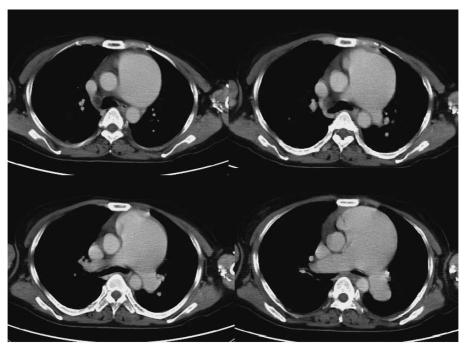


Figure 2. Computed tomography revealed an 6.1cm in diameter in the main pulmonary artery.

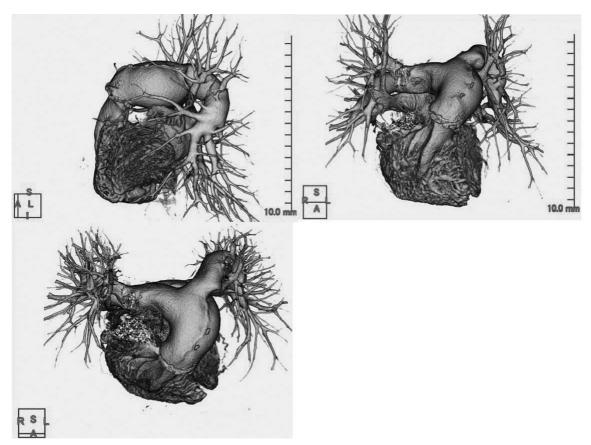


Figure 3. Three-dimensional reconstruction of chest computed tomography. Shown here is surgical repair of pulmonay artery aneurysm.

arterial wall showed no apparent abnormalities, such as cystic necrosis of the tunica media. Warfarin was administered for 3 months post-surgery. The patient postoperatively developed mediastinitis, for which muscle flap closure was performed. She was discharged on postoperative day 99. Nine months after surgery, she was doing well, with no cardiac symptoms or pulmonary arterial dilatation.

## Discussion

PAA is a rare disease (revealed in only 8 cases out of about 110,000 autopsies[1]); therefore, there are no clear guidelines for its assessment or surgical indication for its treatment. Underlying causes include congenital heart disease with intracardiac shunting, the most frequent one being patent ductus arteriosus, followed by ventricular and atrial septal defects, pulmonary hypertension, valvular diseases, connective tissue diseases, infections (bacterial endocarditis, tuberculosis, syphilis), collagen vascular-like diseases, and iatrogenic causes. Aneurysms not caused by these conditions are considered idiopathic. There were no abnormal pathological findings of

the pulmonary artery wall, infections, trauma, collagen-vascular disease, or arteriovenous communication. The aneurysm in this patient is very likely due to dilatation secondary to pulmonary valve insufficiency.

While PAA alone has few symptoms, when complicated by pulmonary valve diseases and cardiac failure, it causes palpitation, fatigue, cough, and dyspnea on exertion. Moreover, as the aneurysm grows, cough, chest pain, and hoarseness may be caused by compression of the bronchi or surrounding organs. In particular, the hemoptysis causes pain, which signifies impending rupture, and surgical repair is recommended as soon as possible.

One study reported that although the prognosis is poor, some aneurysms do not rupture for a long time, with the risk of rupture is low for low-pressure PAAs, and conservative therapy is recommended[2]. However, another study reported that approximately one-third of all PAAs rupture and are often complicated by cardiac failure[3]. Furthermore, some authors suggested monitoring aneurysms <50 mm in diameter without any treatment. However, surgery is recommended for patients with symptomatic and progressively growing aneurysms (50-80 mm in diameter) accompanied by congenital or other underlying diseases or pulmonary hypertension. Additionally, surgery is indicated for aneurysms >80 mm in diameter, regardless of whether they are symptomatic or asymptomatic<sup>[4]</sup>. One study found that a pulmonary arterial pressure of  $\geq$ 50 mmHg, prolonged pulmonary hypertension, pulmonary artery diameter of ≥75 mm, or an annual increase of  $\geq 2$  mm per year in the artery diameter increases the risk of dissection or rupture of PAAs[5], and surgery is recommended in such cases. Furthermore, because vascular wall tension is directly related to the aneurysm's diameter (Laplace's law), the risk of rupture increases even if the arterial pressure is not high. Thus, surgery is often indicated for patients with elevated right ventricular pressure, pulmonary arterial pressure, arterial wall disorders such as dissection, and a PAAs of  $\geq 60$ mm in diameter[6]. There were some of the opinions regarding surgical management; however, there were no official criteria for determining which aneurysms should be surgically managed. Large aneurysms can be unstable. The thinner the vessel wall and the higher the intravascular pressure, the greater the chance of rupture. Our decision to operate on the patient was based on the large size of the lesion (61mm) and severe pulmonary valve insufficiency.

Surgical procedures for arterial reconstruction include pulmonary artery replacement by artificial vascular graft or homograft and aneurysmorrhaphy or arterioplasty, both of which are considered equally effective[7]. If hemodynamics of the aneurysm demonstrate improvement, aneurysmorrhaphy is sufficient[8]. As a simple and shorter procedure, it is particularly effective in cases requiring valve replacement or intracardiac repair, as in our case. However, when performed in patients with a fragile pulmonary arterial wall with apparent abnormalities or with a history of diseases causing these abnormalities, postoperative recurrence is likely. Thus, pulmonary artery replacement should be aggressively selected.

#### Conclusion

We experienced a case of PAA related to pulmonary valve regurgitation, which was successfully treated by pulmonary artery aneurysmorrhaphy and pulmonary valve replacement.

## **Competing interests**

All authors have no competing interests.

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