



TECHNICAL REPORT

Surgical correction of pulmonary artery aneurysm with extrinsic compression of the left main coronary artery: A case report

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ABSTRACT

Pulmonary artery aneurysm (PAA) is a rare disease, with a poorly known natural history, complex diagnosis and may evolve with serious complications, such as compression of adjacent anatomical structures. In some cases, the presence of such complications is what determines the symptoms and is the initial manifestation of the disease. This paper aims to report the case of a patient with typical angina, submitted to cardiac catheterization, which showed, among other lesions, severe left main coronary lesion with characteristics that led to the suspicion of extrinsic compression, which was identified as caused by a PAA. The treatment chosen in this case was surgical, reported concurrently with a literature review that guided the medical team in their decision-making.

Relevance for patients: Surgical correction of PAA may provide resolution of coronary symptoms in affected individuals.

1. Introduction

Pulmonary artery aneurysm (PAA) is a rare condition, first described in 1860, whose prevalence, based on a series of necropsies, is expected to be one in 13,696 individuals [1]. The natural history of the disease is poorly known, and despite its rarity, it may be associated with several causal factors, thus presenting different evolutions [2]. They often have an indolent course and may, in some situations, lead to relating complications, such as compression or rupture of adjacent structures, increasing the morbidity and mortality rate. In such situations, the approach chosen is often surgical.

It is proposed to present an example of this situation in the present case report, evaluated by the research ethics committee of the Goiânia Emergency Hospital, and linked to Plataforma Brasil and receiving its approval under number CAAE: 08 498819.8.0000.0033.

2. Case Report

A 67-year-old female patient looked for cardiology service complaining of precordial pain beginning 3 weeks ago, in tightness, intermittent and associated with respiratory distress on medium exertion. She had hypertension, dyslipidemia,

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congenital heart defects – the most commonly reported are persistent ductus arteriosus and ventricular or atrial septation defects – or presented as a complication of an adjacent cause of pulmonary hypertension, most often due to thromboembolism [2,3].

Congenital heart abnormalities may also cause low-pressure PAA due to high pulmonary flow. Other causes of low-pressure aneurysm include post-stenotic or idiopathic dilatation and vessel wall changes in diseases such as Marfan, Ehlers-Danlos syndromes, and Beçet's disease [3,4]. Pseudoaneurysms may occur due to these same vessel wall abnormalities or due to acquired causes such as adjacent neoplasms and fragility secondary to intravascular procedures such as Swan-Ganz catheter implantation [2,4].

PAA presents a varied spectrum of clinical manifestations, influenced among other factors by aneurysm pressure, size, growth velocity, and adjacent disease [2]. In situations with low pressure and slow growth, the patient may remain asymptomatic for years to decades [5]. In symptomatic cases, it may present with dyspnea, chest pain, hoarseness, palpitations, and syncope [3]. In some situations, clinical manifestations are secondary to the clinical complications of pulmonary aneurysm, including cyanosis, cough, progressive dyspnea, and bronchiectasis in patients with bronchial compression [3]; hemoptysis, rapid onset of circulatory shock in patients with aneurysm rupture [2]; and typical chest pain in individuals with extrinsic coronary compression [6].

In the presence of symptoms compatible with PAA, the suspicion may be increased with the presence of the right ventricular overload on electrocardiogram and hilar enlargement on chest radiography [2]. However, these changes may be absent [7]. Angiography is an important examination defining anatomy and pressure measurement, with caveats for its invasive characteristic and technical difficulty in performing [2,7]. Angiotomography, in turn, presents high accuracy and adequate definition of the anatomy [2]. The diagnosis of extrinsic compression of the LMCA can be a challenge, due to the dynamic characteristic of compression, and therefore, depends on an adequate diagnosis of PAA [6].

The lack of guidelines guiding the best time for surgical approach has been a challenge in cases of PAA; however, literature reviews indicate that pulmonary artery pressure and diameter measurements, as well as its annual growth rate, have been important in determining the risk of aneurysm rupture and thus establish the indication for surgical correction since this complication presents high mortality. Limit values found for surgical planning are 50 mmHg for pulmonary artery pressure, 75 mm diameter, and annual growth rate greater than 2 mm [3].

In individuals with PAA with extrinsic coronary artery compression, determinants of higher risk for ischemia were identified, such as the ratio between the pulmonary artery and aorta diameters greater than 2 and the angle between the LMCA and left sinus of Valsalva $<30^{\text{th}}$ [6,8]. The patient's ischemic assessment by functional testing may be important for the decision of invasive approach [6].

The invasive approach may differ depending on the cause of the pulmonary artery dilation and the degree of cardiac involvement. In clinical situations with low aneurysmal pressure, aneurysmorrhaphy may be the therapy of choice, with a low rate of

intraoperative complications, with the exception that parietal stress may remain [5]. In patients with adequate clinical and anatomical conditions, arterioplasty with or without pulmonary valve replacement (depending on indication) is an advantageous method because it treats both aneurysm and extrinsic compression [9]. Synthetic graft aneurysmectomy, usually Dacron's, associated with pulmonary valve replacement is the procedure of choice in cases with a higher risk of complications, such as pulmonary hypertension [2]. Myocardial revascularization may be performed in these situations, and in some reported cases, stent implantation was the treatment method of choice in patients with severe congenital disease associated with high surgical risk [8,10].

4. Conclusion

PAA is a low prevalence disease that can present as differential diagnosis of coronary obstruction and should be hypothesized in the suspicion of extrinsic compression of the coronary arteries. The proper diagnosis of this clinical condition should be combined with a thorough anatomical evaluation as well as pressure-1 and prognostic evaluation so that the need for a surgical approach is determined.

Disclosure Statement

The authors report no financial relationships or conflicts of interest regarding the content herein.

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