

TECHNICAL REPORT

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

Artur Henrique de Souza¹, Pedro Arthur Ferreira Borges², Victor Eduardo de Almeida e França², Leonardo Veloso do Amaral², Geraldo Paulino Santana Filho³, Jader Bueno Amorim³, Marvyn de Santana do Sacramento⁴, Jefferson Petto^{4,5,6}, Giulliano Gardenghi^{7*}

¹*Department of Cardiovascular Surgery, ENCORE Hospital, Aparecida de Goiânia, GO, Brazil*

²*Department of Clinical Cardiology, ENCORE Hospital, Aparecida de Goiânia, GO, Brazil*

³*Department of Cardiovascular Surgery, Holy House of Mercy of Goiânia, Goiânia, GO, Brazil*

⁴*Program in Physical Therapy, Bahia University Social Center, Salvador, BA, Brazil*

⁵*Bahian School of Medicine and Public Health, Salvador, BA, Brazil*

⁶*Bahia Adventist College, Cachoeira, BA, Brazil*

⁷*Department of Scientific Coordination, ENCORE Hospital, Aparecida de Goiânia, GO, Brazil*

ABSTRACT

Pulmonary artery aneurysm 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 pulmonary artery aneurysm. 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 pulmonary artery aneurysm may provide resolution of coronary symptoms in affected individuals.

Keywords: surgery,aneurysmectomy, angiography, morbidity, angioplasty, stent

Abbreviations: PAA, Pulmonary artery aneurysm; LMCA, Left main coronary artery; Anterior descending

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 individuals¹. The natural history of the disease is poorly known, and despite its rarity, it may be associated with several causal factors, thus presenting different evolutions². 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, linked to Plataforma Brasil and receiving its approval under number CAAE: 08 498819.8.0000.0033.

2. CASE PRESENTATION

A 67-year-old female patient looked for cardiology service complaining of precordial pain beginning three weeks ago, in tightness, intermittent and associated with respiratory distress on medium exertion. She had hypertension, dyslipidemia, overweight, and mild gastritis due to comorbidities and was taking rosuvastatin, acetyl salicylic acid, metoprolol, and lansoprazole. Physical examination without changes, electrocardiogram with sinus rhythm, anterosuperior divisional block and diffusely inverted T-wave. She brought stress echocardiography without segmental changes.

Due to the high risk of the patient, she underwent coronary angiography, and a significant obstructive lesion in the left main coronary artery (LMCA) was identified, without calcification and with a “pencil point” appearance and moderate lesions in the anterior descending (AD) and right coronary artery, no flow changes. During the examination, contrast-injected right-chamber catheterization was performed, with no suspicion of alteration in the right atrium and ventricle, and pulmonary artery trunk enlargement, compatible with aneurysm and suspected coronary trunk compression (Figure 1). Right chamber pressures were verified, with pulmonary trunk pressure of 35 mmHg.

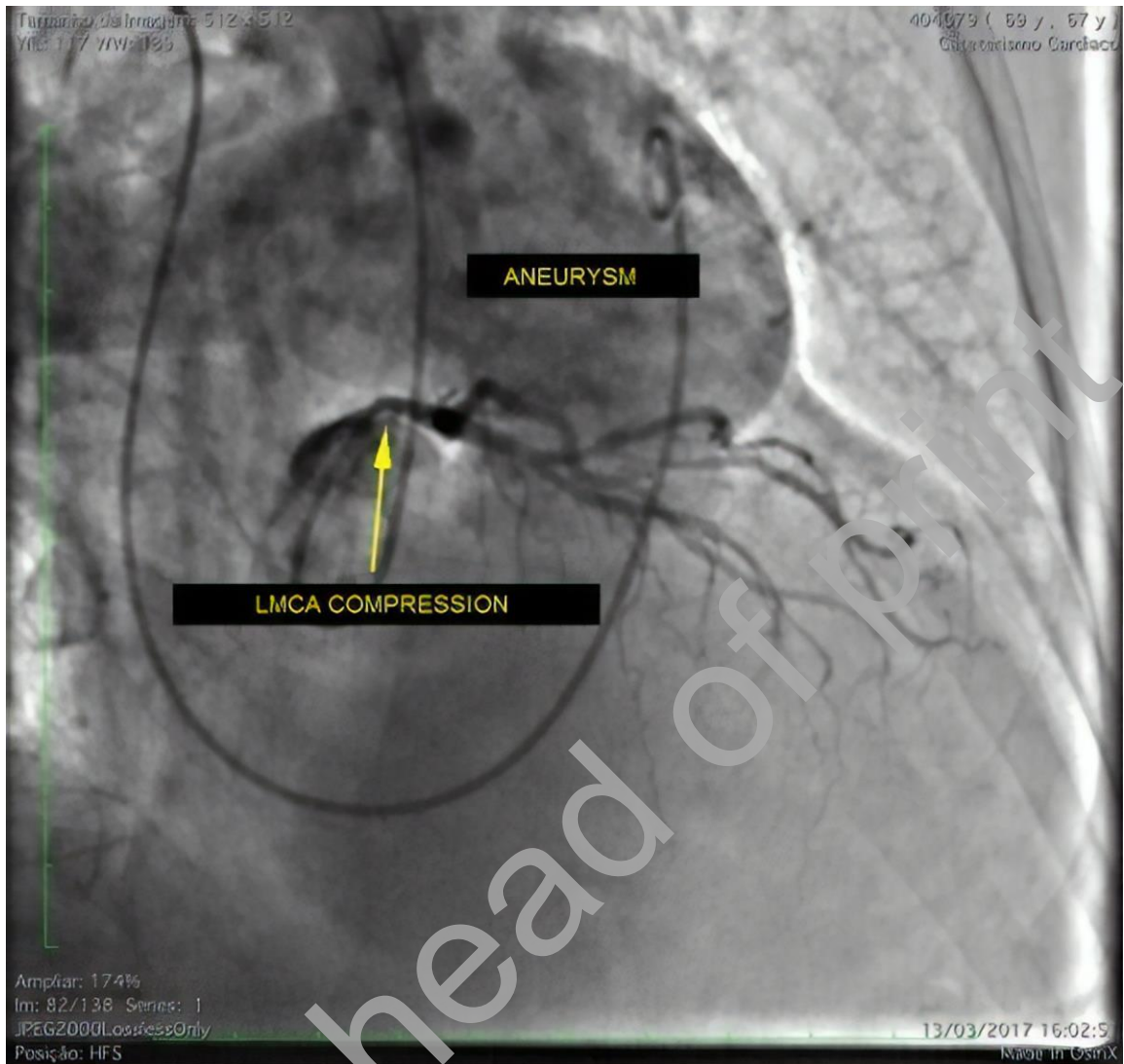


Figure 1. Angiography showing the pulmonary artery trunk, compatible with aneurysm and suspected coronary main compression.

The patient underwent coronary angiography, identifying a pulmonary artery trunk aneurysm measuring 50 mm and finding extrinsic compression of the LMCA (Figure 2).

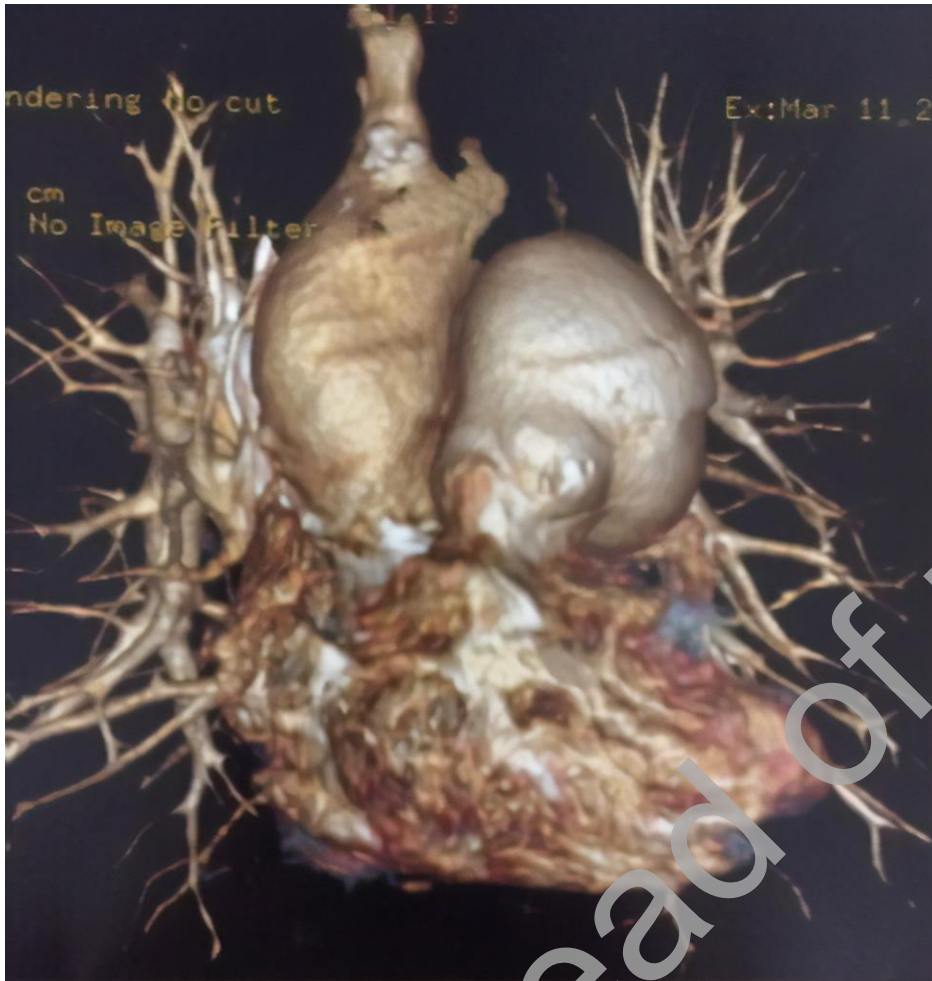


Figure 2. Coronary angiotomography showing pulmonary main artery aneurysm measuring 50 mm, identifying extrinsic compression of left main coronary artery.

In the present case, surgical correction of the pulmonary artery trunk aneurysm was indicated. The procedure was performed by median sternotomy and after pericardiotomy and full heparinization, cardiopulmonary bypass was started with the venous return through the right atrium and arterial return in the distal descending aorta under normothermia. After aortic clamping, a cardioplegic solution (modified St. Thomas) was performed and a complete resection of the pulmonary artery trunk was performed until the confluence of the right and left pulmonary branches with Dacron tube number 28 (Figure 3). After removal of the aortic forceps and return of the heartbeat, hemostasis was reviewed and subsequently closed by planes. The postoperative evolution was satisfactory, and the patient was discharged on the fifth postoperative day, in good general condition, with improvement of symptoms related to compression of the LMCA.

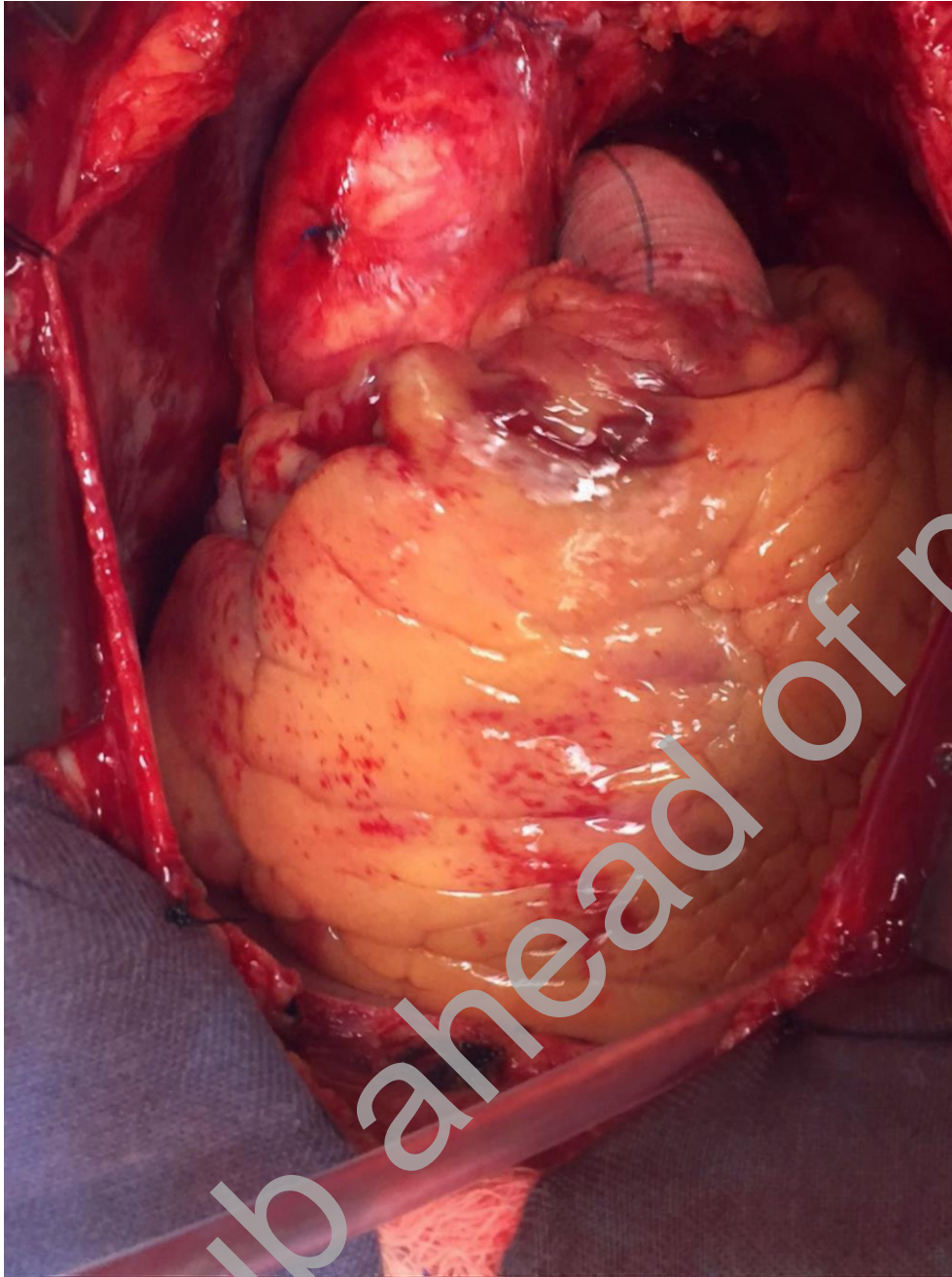


Figure 3. Complete resection of the pulmonary main artery up to the confluence of the right and left pulmonary branches with Dacron tube implantation.

In the present case, surgical correction was chosen when establishing the anatomical diagnosis, finding a pulmonary artery base pressure measurement of 35 mmHg and the size of the aneurysm, since its growth determined extrinsic compression of the LMCA to an extent sufficient to determine anginal symptoms and ischemic alteration in functional cardiac test. Coronary angiography revealed a lesion in the middle third of AD coronary artery, without significant obstruction and without indication of approach at the time.

In outpatient follow-up, symptoms recurred, and the patient underwent a new catheterization 16 months after the surgery, when an increase in the degree of obstruction in AD was identified, treated with angioplasty and implantation of a pharmacological stent and angiographically confirmed LMCA patency when compared to preoperative control.

3. DISCUSSION

Although it is a rare disease, PAA has different causal factors. High-pressure aneurysms may be idiopathic, associated with 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 disease². In situations with low pressure and slow growth, the patient may remain asymptomatic for years to decades⁵. In symptomatic cases, it may present with dyspnea, chest pain, hoarseness, palpitations and syncope³. 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 compression³; hemoptysis, rapid onset of circulatory shock in patients with aneurysm rupture²; and typical chest pain in individuals with extrinsic coronary compression⁶.

In the presence of symptoms compatible with PAA, the suspicion may be increased with the presence of right ventricular overload on electrocardiogram and hilar enlargement on chest radiography². However, these changes may be absent⁷. Angiography is an important exam 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 anatomy². The diagnosis of extrinsic compression of the TCS can be a challenge, because of the dynamic characteristic of compression, and therefore, depends on an adequate diagnosis of PAA⁶.

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 50mmHg for pulmonary artery pressure, 75mm diameter and annual growth rate greater than 2mm³.

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 less than 30th^{6,8}. The patient's ischemic assessment by functional testing may be important for the decision of invasive approach⁶.

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 remain⁵. 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 compression⁹. 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 hypertension². 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. CONCLUSIONS:

PAA is a low prevalence disease that can present as differential diagnosis 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.

REFERENCES

1. Deterling RA Jr, Clagett OT. Aneurysm of the pulmonary artery: review of the literature and report of a case. *Am Heart J* 1947;34:471–499.
2. Kreibich M, Siepe M, Kroll J, Hohn R, Grohmann J, Beyersdorf F. Aneurysms of the pulmonary artery. *Circulation* 2015;131:310–316.
3. Duijnhouwer AL, Navarese EP, Van Dijk APJ, Loeyls B, Roos-Hesselink JW, De Boer MJ. Aneurysm of the pulmonary artery, a systematic review and critical analysis of current literature. *Congenit Heart Dis* 2016;11:102–109.
4. Park HS, Chamarthy MR, Lamas D, Saboo SS, Sutphin PD, Kalva SP. Pulmonary artery aneurysms: diagnosis & endovascular therapy. *Cardiovasc Diagn Ther* 2018;8:350-361.
5. Veldtman GR, Dearani JA, Warnes CA. Low pressure giant pulmonary artery aneurysms in the adult: natural history and management strategies. *Heart* 2003;89:1067–1070.
6. Kajita LJ, Martinez EE, Ambrose JA, Lemos PA, Esteves A, Nogueira da Gama M, Jatene AD, Ramires JA. Extrinsic compression of the left main coronary artery by a dilated pulmonary artery: clinical, angiographic, and hemodynamic determinants. *Catheter Cardiovasc Interv* 2001;52:49–54.
7. Tsui EY, Cheung YK, Chow L, Chau LF, Yu SK, Chan JH. Idiopathic pulmonary artery aneurysm: digital subtraction pulmonary angiography grossly underestimates the size of the aneurysm. *Clin Imaging* 2001;25:178-180.

8. Dodd JD, Maree A, Palacios I, de Moor MM, Mooyaart EA, Shapiro MD, et al. Images in cardiovascular medicine. Left main coronary artery compression syndrome: evaluation with 64-slice cardiac multidetector computed tomography. *Circulation* 2007;115:e7-8.
9. Yeh DD, Ghoshhajra B, Inglessis-Azuaje I, MacGillivray T, Liberthson R, Bhatt AB. Massive pulmonary artery aneurysm causing left main coronary artery compression in the absence of pulmonary hypertension. *Tex Heart Inst J* 2015;42:465–467.
10. Ikegami R, Ozaki K, Ozawa T, Hirono S, Ito M, Minamino T. Percutaneous Coronary Intervention for a Patient with Left Main Coronary Compression Syndrome. *Intern Med.* 2018;57:1421-1424.

Epub ahead of print