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SCIENTIFIC ARTICLE - CASE REPORT

COMBINED PERCUTANEOUS TREATMENT OF GIANT ARTERIOVENOUS MALFORMATION AND COMPLEX CORONARY LESIONS IN A HIGH-RISK SURGICAL PATIENT: A CASE REPORT

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ABSTRACT

Coronary artery fistulas (CAF) are considered rare congenital anomalies, characterized by direct communications between the coronary arteries and cardiac chambers or great vessels. Their prevalence is estimated at 0.2% to 0.4% of congenital heart defects. Their etiology is related to abnormalities in embryonic development. The right coronary artery (RCA) and the left anterior descending artery (LAD) are the most common sites of origin. Clinical presentation is variable, ranging from asymptomatic cases to symptoms such as fatigue, dyspnea, palpitations, and chest pain (due to ischemia). Diagnosis is established through imaging methods such as echocardiography, cardiac computed tomography angiography, and coronary angiography. Currently, there are no standardized therapeutic guidelines for the management of CAF due to the rarity of cases. This article aims to report a case of a patient with CAF and complex coronary lesions who underwent coronary angioplasty and fistula embolization concomitantly.

Keywords: Fistula, Embryonic development, Left anterior descending artery, Right coronary artery.

INTRODUCTION

Perinatal Coronary artery fistulas (CAF) are rare congenital anomalies characterized by direct communications between one or more coronary arteries and cardiac chambers or adjacent great vessels. The prevalence of CAF is estimated at 0.2% to 0.4% of all congenital heart defects[1]. Among the various forms of CAF, fistulas from the coronary artery to the pulmonary trunk (CAPF) are particularly uncommon, accounting for approximately 17% of CAF cases.[2]

The etiology of CAPF is predominantly attributed to abnormalities in the embryonic development process, specifically the persistence of embryonic myocardial sinusoids or incomplete involution of branches of the pulmonary sinus. The right coronary artery (RCA) and the left anterior descending artery (LAD) are the most frequently identified origins of these fistulas.[1]

The clinical presentation of CAPF is highly variable, ranging from asymptomatic cases to nonspecific manifestations that resemble other heart diseases. When present, symptoms are

intrinsically related to the magnitude of blood flow shunting (left-to-right shunt) and the diameter of the fistula. Symptoms may include fatigue, exertional dyspnea, palpitations, chest pain (including myocardial ischemia due to the "coronary steal" phenomenon), and syncope.[1] It is noteworthy that syncope is an atypical manifestation and is rarely considered in the differential diagnosis of vascular malformations. On physical examination, most patients may not present significant findings; however, some may exhibit a continuous murmur suggestive of patent ductus arteriosus.

Regarding complementary examinations, the electrocardiogram rarely shows specific abnormalities. The diagnosis is primarily established through imaging methods such as echocardiography and cardiac computed tomography angiography. Coronary angiography is considered the gold standard for diagnostic confirmation and anatomical delineation of CAPF[1].

Currently, there are no standardized therapeutic guidelines for the management of CAPF.[3] The scarcity of reported cases makes it difficult to establish universal management strategies. Therefore, the description of new cases significantly contributes to improving knowledge about the pathophysiology, clinical course, and potential complications of this condition, as well as assisting in the clarification of complex differential diagnoses.

The aim of this article is to report the case of a patient with complex obstructive coronary artery disease and multiple giant coronary fistulas to the pulmonary artery trunk, treated with coronary angioplasty associated with concomitant fistula embolization.

CASE REPORT

A 66-year-old female patient, with a history of systemic arterial hypertension, insulin-dependent diabetes mellitus, dyslipidemia, stage IV chronic kidney disease (non-dialysis), and pulmonary hypertension, was evaluated.

The initial diagnostic assessment, including myocardial perfusion scintigraphy, revealed significant ischemia (approximately 21%), with hypoperfusion involving the anterior, septal, and apical regions.

She underwent cardiac catheterization (CATH), which revealed heavily calcified lesions and significant obstructions: RCA: 90% proximal; LAD: 95% proximal and 90% in the mid segment (Figures 1A and 1C); left circumflex artery (LCx): 90% proximal; posterior descending artery (PDA): 80% in the mid segment. In addition, high-flow fistulas originating from the proximal segments of the LAD and RCA to the pulmonary trunk were identified (Figures 1B and 1D). Regarding pulmonary hypertension (pulmonary artery systolic pressure of 60 mmHg on CATH) and coronary-cameral fistulas, manometry performed during CATH revealed significant pulmonary arterial hypertension, and the presence of coronary-cameral fistulas was confirmed.

Given the patient's high surgical risk, the Heart Team decided on percutaneous treatment with coronary stenting and fistula embolization. The LAD was prepared with rotational atherectomy using a 1.5 mm burr, associated with a cutting balloon (Wolverine – Boston Scientific 3.5 × 15 mm) (Figure 1C and Figures 2A and 2B), followed by implantation of two everolimus-eluting stents (4.0 × 23 mm and 3.5 × 32 mm). The fistula originating from the LAD was embolized with 20 microcoils (Figure 2C).

In the same procedure, the RCA was selectively catheterized, and two guidewires were positioned: one along the fistula tract (Guidezilla 6F microcatheter) and another along the native coronary pathway (XB 3.5 6F guiding catheter). A 4 mm Amplatzer® AVP2 vascular plug was deployed in the proximal portion of the fistula, leaving treatment of the coronary lesion for a second stage, when complete occlusion of the malformation would be confirmed or not (Figure 2D).

Thirty days later, the patient returned for RCA stent implantation. Pre-intervention angiography

demonstrated unsuccessful fistula occlusion with a single device, with persistent high flow to the pulmonary trunk. We opted to treat the mid segment of the RCA, again using rotational atherectomy, followed by implantation of an initial stent in the mid segment (4.0 × 38 mm) to ensure adequate coronary flow.

Fistula embolization was then performed by crossing the previously implanted proximal plug with a guidewire (Hornet 0.14"), advancing the Guidezilla 6F microcatheter through the mesh of the plug into the mid segment of the fistula body, and deploying 8 microcoils, achieving complete occlusion confirmed by angiography. The microcatheter was then withdrawn to the proximal segment between the plug and the origin of the fistula, where 2 additional microcoils were released. Finally, a second stent (4.0 × 23 mm) was deployed in the proximal RCA with successful outcome (Figure 2D).

Reevaluation of the left coronary artery demonstrated complete occlusion of the arteriovenous fistula, with the LAD showing excellent flow and caliber. The patient remained hospitalized in the Intensive Care Unit after the procedure and was discharged the following day for outpatient follow-up.

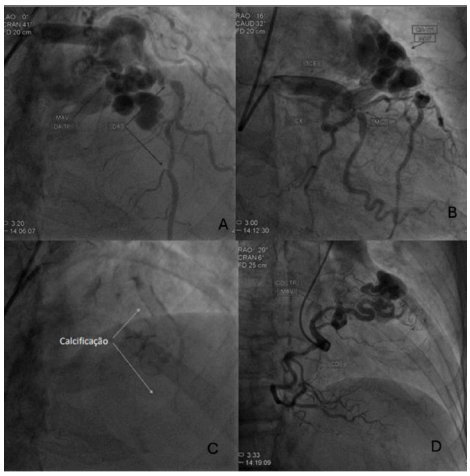

Figure 1: Angiographic images: 1A: Severe atherosclerotic coronary lesion in the left anterior descending artery. 1B: Fistula from the left anterior descending artery to the pulmonary artery trunk. 1C: Calcified atherosclerotic lesion in the left anterior descending artery. 1D: Fistula from the right coronary artery to the pulmonary artery trunk.

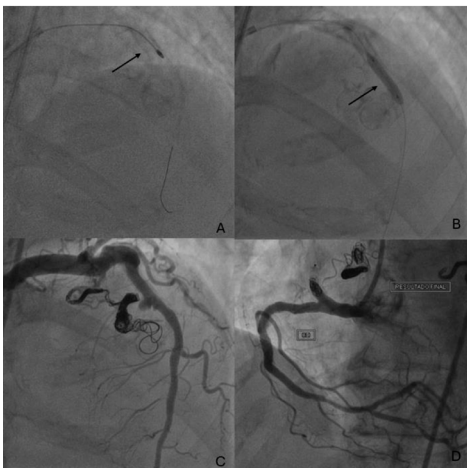

Figure 1: Angiographic images: 2A: Rotational atherectomy in an atherosclerotic lesion of the left anterior descending artery. 2B: Cutting balloon in the left anterior descending artery. 2C: Final result of left anterior descending artery angioplasty and fistula occlusion. 2D: Final result of right coronary artery angioplasty and fistula occlusion.

DISCUSSION

Coronary artery fistula to the pulmonary artery (CAPF) is a rare subtype, accounting for approximately 17% of all CAF cases.[1] Its incidence in the general population is estimated at 0.002%, with most cases being congenital. However, it may also be acquired due to trauma, iatrogenic coronary interventions, acute myocardial infarction, hypertrophic or dilated cardiomyopathy, and malignancy.[4]

Many patients with CAF are asymptomatic, especially those with small and hemodynamically insignificant shunts.[3] However, the frequency of symptoms and complications increases with age, and the presentation of large fistulas in older individuals is extremely rare.[4] The most common symptoms include angina (chest pain), dyspnea (shortness of breath), palpitations, dizziness, and syncope.[2] Angina may result from coronary artery disease (CAD) or from the coronary steal phenomenon.[5] Coronary steal occurs when blood flow is diverted to a low-resistance receiving chamber (such as the pulmonary artery), leading to ischemia in myocardial regions with higher vascular resistance.

In a case reported by Saeed et al., myocardial ischemia resulting from coronary steal secondary to a fistula between the LAD and the pulmonary artery led to a non-ST-elevation myocardial infarction in a young patient.[4] In our case report, the patient also presented significant ischemia on myocardial scintigraphy; however, with a multifactorial etiology related to the presence of obstructive coronary artery disease and the fistula.

The management of CAF remains controversial due to the rarity of cases and the lack of standardized treatment guidelines. The choice of therapy depends on the size of the fistula, its anatomical location, the patient's clinical presentation, and the presence of documented coronary steal.[4] Therapeutic options include clinical management, surgical treatment, or transcatheter embolization.[3]

In a second case also reported by Saeed et al., the patient showed symptom improvement with clinical treatment alone, without the need for surgical or endovascular intervention[4]. In contrast, in our case, the presence of significant ischemia associated with obstructive coronary artery disease precluded conservative management, leading to the decision to perform coronary angioplasty for the coronary obstruction and embolization with coils and a vascular plug due to the large-volume fistula in the LAD and RCA.

Surgical treatment remains the traditional gold standard and is considered a safe and effective option. It is performed via median sternotomy, with or without cardiopulmonary bypass.[3] Surgical options include epicardial ligation, transection with closure of the opening, and drainage.[3] The presence of a fistula requiring treatment demands evaluation by an experienced team, including cardiologists with expertise in congenital heart disease and a cardiac surgeon, to determine the best therapeutic approach.[6]

Direct comparative studies between surgical and endovascular treatment are still lacking, and both are considered viable therapeutic options. Percutaneous closure is preferred in cases of proximally originating fistulas with a single drainage site, whereas surgical treatment is generally reserved for distal fistulas and those with multiple drainage sites.[4] In our case, given the presence of associated obstructive atherosclerotic coronary disease, the expertise of the Heart Team in percutaneous treatment of coronary and structural heart disease, and the patient's high surgical risk, a simultaneous percutaneous approach for both conditions was chosen.

CONCLUSION

Coronary artery fistulas to the pulmonary artery (CAPF) may represent a differential diagnosis in patients with acute or chronic coronary syndromes, mainly due to the coronary steal phenomenon, especially when associated with significant atherosclerotic disease. Mastery of multiple percutaneous intervention techniques

(used in endovascular surgery and neurointervention) allows, in selected cases, complete resolution of both the malformation and the atherosclerotic disease, particularly in patients with high surgical risk.

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EDITORIAL AND REVIEW

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