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Bilateral coronary-pulmonary artery fistulas associated with coronary artery stenosis

Abstract

Coronary artery venous fistulas (CAVFs) are rare congenital anomalies often detected incidentally during routine coronary angiography. While asymptomatic CAVFs are usually managed conservatively due to their potential to regress or close spontaneously, symptomatic CAVFs require intervention. This report presents the case of a 67-year-old female patient with a history of hypertension, type 2 diabetes, and hypercholesterolemia, who presented with chest pain and exertional dyspnea. Routine angiography revealed bilateral CAVFs with fistulas originating from both the left and right coronary arteries, in addition to critical stenoses in the left anterior descending artery and the circumflex artery. The patient underwent CAVFs closure surgery combined with coronary artery bypass grafting (CABG). Postoperatively, all cardiac symptoms were completely resolved. This case highlights that CAVFs should also be considered as a rare cause of ischemic heart disease and demonstrates the effectiveness of surgical intervention in the treatment of symptomatic cases.

Keywords: Coronary artery disease, coronary pulmonary arteriovenous fistula, surgical treatment

INTRODUCTION

Coronary arteriovenous fistulas (CAVFs) are characterized by abnormal connections between coronary vessels and either the cardiac chambers or great vessels. It has been reported that CAVFs were first described by Krauss in 1865 (1). In the general population, the incidence is approximately 0.002% (2). Although these anomalies are infrequent, they represent the most common hemodynamically significant abnormality of the coronary arteries. The prevalence of CAVFs has been documented in 0.15–5.3% of cases referred for routine coronary angiograms or catheterizations (3). We present a case of bilateral coronary artery venous fistulas detected during routine angiography in the evaluation of acute coronary syndrome, subsequently treated with coronary artery bypass graft surgery and CAVFs closure.

CASE REPORT

A 67-year-old female patient presented to the hospital with complaints of chest pain and dyspnea upon minimal exertion (NYHA Class III). The patient's medical history was significant for

hypertension, but no family history was identified. Transthoracic echocardiography revealed a fibrotic mitral valve with minimal mitral regurgitation and reduced global systolic function of the left ventricular myocardium due to apicolateral hypokinesia, with an ejection fraction of 50%. Coronary angiography demonstrated extensive coronary lesions, including critical stenoses of the left anterior descending (LAD) artery (70%) and the circumflex artery (Cx) (80%), as well as a fistulous branch originating from the proximal third of the LAD (Figure 1). However, the exact anatomical drainage of this branch could not be determined in coronary angiography. Subsequent coronary CT angiography revealed a fistulous structure originating from the proximal third of the LAD and draining into the pulmonary artery. In addition, CT angiography revealed a second coronary fistula, which had not been observed in the coronary angiography. The second fistula was located between the highest point of the right coronary artery (RCA) orifice and the pulmonary artery (Figure 2). The patient underwent coronary bypass surgery via median sternotomy. Upon opening the pericardium, the fistulous branch originating from the proximal third of the LAD and draining

into the pulmonary artery was clearly observed. Additionally, the fistula originating near the right coronary artery and draining into the pulmonary artery was also visualized (Figure 2). Bypass grafting was performed on the LAD and Cx arteries. Subsequently, both fistulas were closed at their drainage points into the pulmonary artery using ligaclips. The patient had an uneventful postoperative course and was discharged on the 6th postoperative day.

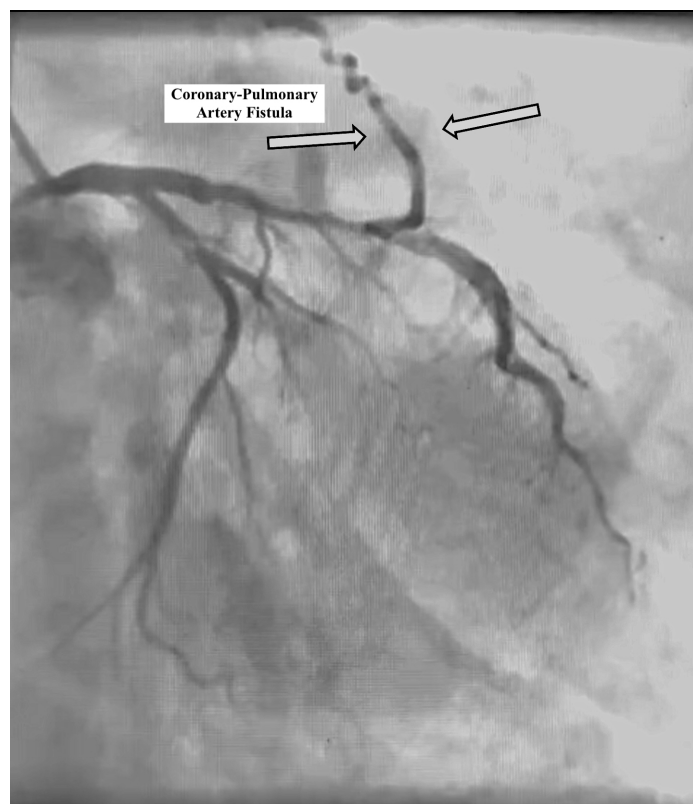


Figure 1. Coronary angiogram view of the coronary-pulmonary artery fistula

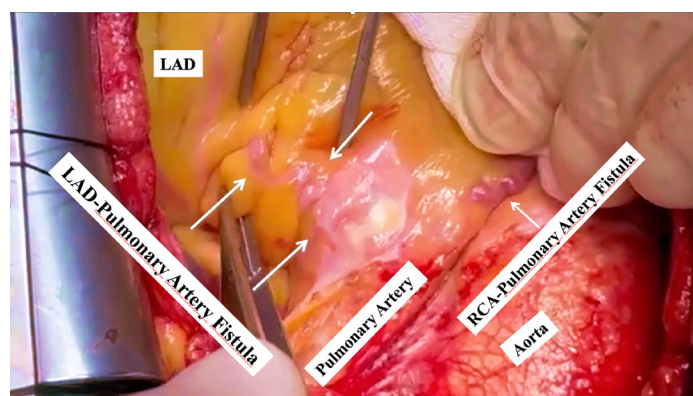


Figure 2. Operative view of the coronary-pulmonary artery fistula.

DISCUSSION

CAVFs are extremely rare anomalies, occurring in 0.002% of the general population and accounting for 0.4% of all cardiac malformations (2). CAVFs have been reported to originate most frequently from the RCA (50%), followed by the LAD (25%)

and the Cx artery (18.3%), with an equal frequency of drainage into either the left or right pulmonary arteries. Bilateral fistulas are much rarer, accounting for only 5% of all coronary artery fistulas (4,5). The presence of two CAVFs simultaneously in our case is noteworthy.

Most CAVFs are congenital; however, CAVFs can also develop following congenital heart surgeries, myocardial biopsy, coronary angioplasty, and coronary bypass surgery (5). The formation of congenital CAVFs is often explained by Hackensellner's involution persistence hypothesis. According to this theory, the failure of the normal capillary network to develop between the coronary arteries and the great vessels or ventricular chambers during embryogenesis is proposed as the cause of major CAVFs (6,7). Coronary angiography is the standard imaging method for evaluating the coronary arteries. In our case, however, we observed that not all CAVFs could be detected by coronary angiography alone. Unlike normal coronary anatomy, CAVFs originating directly from the aorta cannot be easily detected by coronary angiography. Therefore, we believe that in patients suspected of having CAVFs, performing CT angiography as an additional standard procedure is necessary both for visualizing the CAVFs and identifying their drainage sites.

Patients with CAVFs do not exhibit definitive symptoms. However, most symptoms, such as exertional dyspnea and angina, typically appear in older age groups (2). Our patient presented with chest pain at the age of 67. Chest pain is generally attributed to blood steal from the high-pressure coronary artery system to the low-pressure pulmonary artery system (6). Although no direct relationship between CAVFs and coronary artery lesions has been established, our patient had both CAVFs and coronary stenoses. Postoperatively, the chest pain completely resolved, but it is not possible to determine whether this was due to coronary bypass surgery or the closure of the CAVFs.

Unlike small-diameter asymptomatic CAVFs, there is a consensus that all symptomatic CAVFs should be closed. It has been reported that asymptomatic, small-diameter CAVFs can regress and close spontaneously (7). In our case, we were unable to determine the extent of blood steal by the CAVFs from the coronary arteries. The CAVFs flow volume can be measured preoperatively in the angiography laboratory or intraoperatively using ultrasonography. However, due to the lack of technical equipment, we were unable to perform this assessment in our clinic. Nevertheless, during coronary angiography, the CAVFs appeared to have reached a diameter similar to that of the LAD, suggesting a high-flow fistula. The selection of the treatment modality for CAVFs, whether surgical or transcatheter, depends on the expertise of the treating physicians. The decision-making process for fistula management requires a thorough evaluation of several factors, including: The anatomical origin and size of the CAVFs, patient's symptoms, presence of concomitant conditions such as angina, heart failure, endocarditis, or rupture, and necessity for an additional invasive procedure (8). In our patient, ischemic symptoms were prominent, and surgical intervention

was planned to address both coronary revascularization and CAVFs closure.

CONCLUSION

This case underscores that, while rare, CAVFs should be considered as a potential etiological factor in patients presenting with symptoms indicative of coronary artery stenosis. Moreover, it highlights that such cases can be successfully managed through appropriate surgical intervention.

Conflict of Interests: The authors declare that there are no conflict of interests.

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Patient Informed Consent: A written informed consent form was obtained from the patient.

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