



Case Report

Bilateral communicating coronary arteriovenous fistulas presenting with recurrent angina

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ABSTRACT

Coronary fistula is a rare disorder. Early diagnosis is difficult because angina caused by stenosis secondary to coronary atheroma is very similar to the coronary steal phenomenon. We report the case of a 51-year-old man who presented with recurrent angina over an 8-year period in whom bilateral coronary fistulas were detected by conventional coronary angiography. Sixty-four-slice multidetector computed tomography (MDCT) showed bilateral coronary fistulas merging to form an aneurysm before entering the pulmonary artery. However, thallium²⁰¹ single-photon emission computed tomographic (SPECT) myocardial perfusion imaging showed only inferior apical ischemia. The combined use of these techniques showed the detailed anatomy of the fistula and the degree of myocardial ischemia for clinical planning. Both MDCT and SPECT are important diagnostic tools and should be considered in the clinical evaluation of bilateral coronary fistulas.

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1. Introduction

Coronary artery fistulas (CAFs) are congenital or acquired anomalies in which a coronary artery is connected with either a cardiac chamber or a great vessel. These coronary anomalies can cause myocardial ischemia owing to the coronary steal phenomenon. Studies have shown that the incidence of CAF is about 0.3–0.8% in coronary anomalies and bilateral CAFs are even more rare, representing only 5% of the total number of coronary fistula cases [1]. The clinical presentation and imaging for patients with CAFs can vary considerably, making early detection difficult. In this article, we report a patient presenting with recurrent angina who received thallium²⁰¹ single-photon emission computed tomographic (SPECT) myocardial perfusion imaging, coronary angiography and 64-slice multidetector computed tomography (MDCT), allowing the identification of a rare case of bilateral CAFs communicating to form an aneurysm.

2. Case report

A 51-year-old man presented to our hospital with recurrent angina over an 8-year period. The results of his physical examination, laboratory tests and chest radiography showed no abnormalities. Resting and treadmill electrocardiograms showed no obvious ischemic changes. SPECT showed a reversible perfusion defect over the inferior wall and part of the apical wall. Coronary angiography revealed a fistula arising from the left anterior descending (LAD) artery and another fistula arising from the right coronary artery (RCA). Subsequent contrast injections showed both fistulas entering the pulmonary artery (Fig. 1A and B). The three coronary arteries were free of significant atherosclerotic lesions. Sixty-four-slice MDCT showed that the two fistulas from the LAD and RCA merged to form an aneurysm before entering the pulmonary artery (Fig. 2A). Surgical closure of the CAFs was recommended because of inferior apical ischemia caused by the coronary steal phenomenon (Fig. 2B). However, our patient declined surgery.

3. Discussion

Coronary fistulas are coronary abnormalities in which blood drains into a cardiac chamber or other structure directly instead of

Conflict of interest: none.

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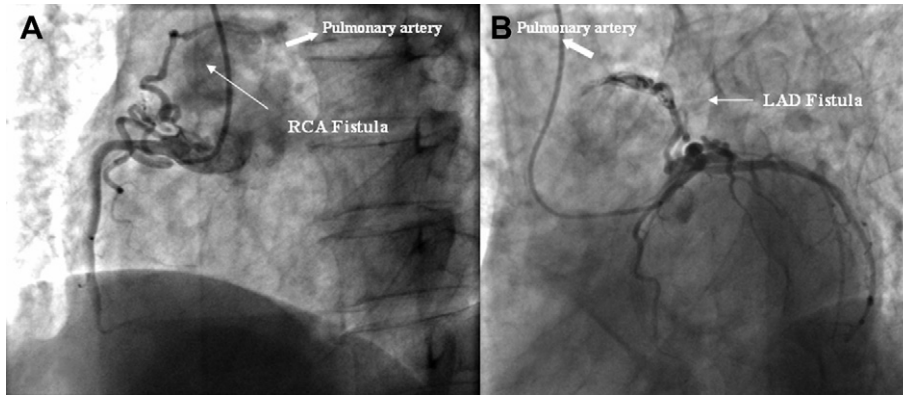


Fig. 1. Coronary angiography: (A) right coronary angiography in the left anterior oblique projection showing a fistula (white arrow) arising from the proximal RCA and draining into the pulmonary artery; (B) left coronary angiography in the left anterior oblique projection showing another fistula (white arrow) arising from the LAD artery and terminating in the pulmonary artery. LCA = left coronary artery; RCA = right coronary artery.

passing through the myocardial capillary network. They can be either congenital or acquired. Myocardial ischemia, heart failure, and bacterial endocarditis are known complications. Treatment including surgical closure and catheter-based closure is recommended for patients with symptoms of heart failure or myocardial ischemia or asymptomatic patients with high-flow shunting. Our patient had recurrent angina caused by the coronary steal phenomenon owing to shunting of the CAFs. SPECT results suggested an underlying myocardial ischemia, and bilateral coronary fistulas were found unexpectedly during conventional coronary angiography. MDCT imaging revealed that the bilateral coronary fistulas joined to form an aneurysm before entering the pulmonary artery. Coronary fistulas usually drain into the right ventricle, right atrium, pulmonary artery, left ventricle, left atrium, and coronary sinus. Fifty-six percent of bilateral fistulas and 17% of unilateral fistulas drain into the pulmonary artery [2]. After a brief review of the English language literature using “bilateral coronary fistula” as a keyword in the PubMed database, we found only three cases of bilateral communicating coronary fistulas [3–5]. All three cases were detected by MDCT and reported after the year 2006. Note that in conventional sequential coronary angiography, a communication

between bilateral CAFs cannot be detected unless contrast material is injected into the RCA and LAD simultaneously. Conversely, MDCT of the heart, a relatively new but increasingly popular technique for coronary assessment, enables detailed evaluation of the anatomy of fistulas and detection of communication between them. In recent years, the clinical value of MDCT for investigating coronary anomalies has been emphasized [6]. Thus, the use of MDCT should be considered in the evaluation of patients with bilateral coronary fistulas discovered by angiography.

The role of SPECT studies in CAFs is unclear. A correlation between reversible perfusion defects owing to the coronary steal phenomenon and the territory of a single coronary fistula has been reported [7]. However, from the literature studies only four patients with bilateral CAFs underwent SPECT; in two of these cases SPECT did not reveal perfusion defects [2,3] and in the remaining two cases it showed compatible perfusion defects [8,9]. In our patient, SPECT showed reversible perfusion defects over the inferior wall and part of the apical wall, which were correlated with the perfusion area of the RCA. The patient’s large LAD fistula did not cause significant anterior ischemia according to the results of SPECT. It is possible that the two shunting forces from the LAD and RCA could

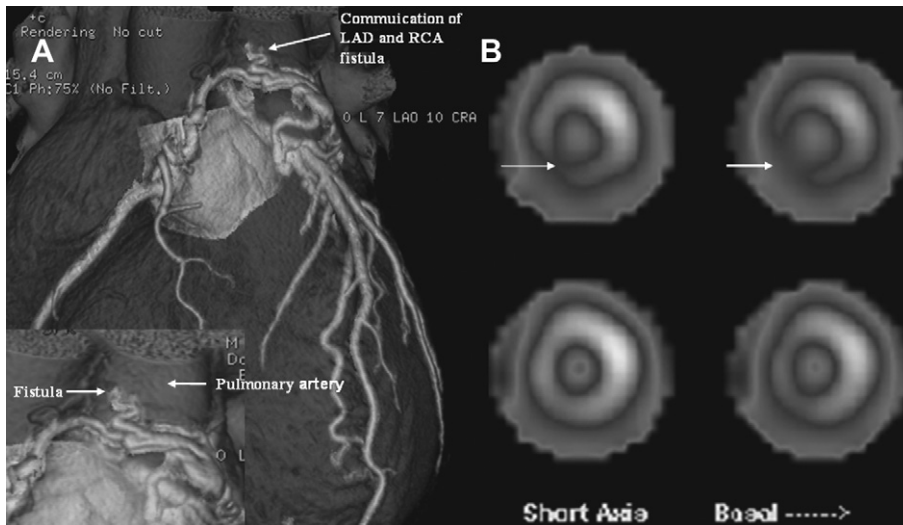


Fig. 2. MDCT and SPECT: (A) MDCT image demonstrates bilateral coronary fistulas from the LAD and RCA to the pulmonary artery merging to form an aneurysm (white arrow) before entering the pulmonary artery (inset bottom left: pulmonary artery and fistula); (B) myocardial perfusion imaging studies showing a reversible perfusion defect (white arrow) over the inferior wall and part of the apical wall. LCA = left coronary artery; MDCT = 64-slice multidetector computed tomography; RCA = right coronary artery; SPECT = thallium²⁰¹ single-photon emission computed tomography.

have interfered with each other causing formation of the aneurysm. Our case showed that the functional consequences of bilateral coronary fistulas and the direction of coronary steal can be difficult to predict if the two fistulas are communicating. Therefore, functional assessment of myocardial ischemia with SPECT is important for surgical planning.

In summary, it is rare to find coronary fistulas that communicate to form an aneurysm. Appropriate imaging techniques are vital for accurate clinical planning. We have reported our experience using imaging methods to assess this rare presentation. MDCT is a powerful diagnostic tool to show the detailed anatomy of rare coronary anomalies and SPECT can be used to evaluate the severity of myocardial ischemia. These two imaging techniques should both be considered in evaluating patients with bilateral CAFs.

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