

Late-term Post-operative Recanalization of a Fistula between the Coronary Artery and Bronchial Vein

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A coronary artery fistula is a rare congenital anomaly that creates a direct link between the coronary artery and the heart chambers or other vascular structures. It is often identified coincidentally during diagnostic coronary angiography and may cause coronary steal syndrome. We report on a 54-year-old man with myocardial ischemia who had coronary angiography and a recanalized fistula was detected during the procedure.

Coronary artery fistulas (CAFs) are abnormal links between epicardial coronary arteries and other cardiac chambers, large veins or other vascular structures. Most CAFs are small and are identified coincidentally during coronary angiography. However, large fistulas can cause myocardial ischemia and angina due to coronary steal syndrome. CAFs may cause several cardiac problems such as acute myocardial infarction, sudden cardiac death, congestive heart failure, endocarditis, stroke, arrhythmia, and coronary aneurysm rupture. CAFs were found at a frequency of 0.18 % in a series of 33,000 patients who had coronary angiography.¹ In a Turkish series, this frequency was found to be approximately 0.08 %.²

We present a case report on a man with a CAF that caused myocardial ischemia between the coronary artery and the bronchial vein which was surgically ligated five years ago and recanalized in the late term. It has not been reported in the literature so far.

Case

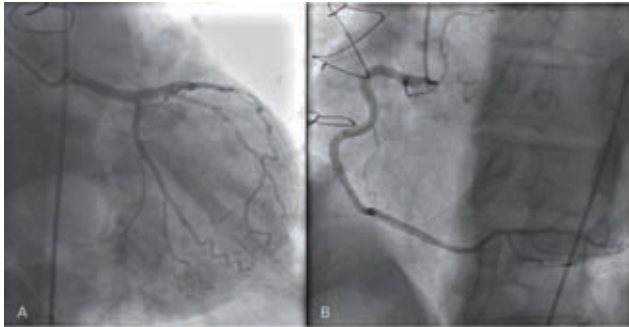
A 54-year-old man presented to our clinic with chest pain upon exertion. The patient had hypertension and non-insulin-dependent diabetes mellitus. He had

coronary angiography five years ago because of angina pectoris. The coronary angiography was normal but a congenital CAF between the right coronary artery (RCA) and the bronchial vein was found. It was surgically closed because it was causing ischemia. The fistula between the RCA and the bronchial vein was isolated parallel to the non-coronary sinus annulus of the aorta and the right atrium chamber. The fistula was tied off with nylon double tape and closed with the placement of a 5/0 transfixion suture in the center. The patient was discharged with a stable condition. During examination, electrocardiography was normal but the exercise test was positive. Echocardiography revealed normal left ventricle systolic function, and slight dilation (4.2 cm) of the aortic root. Coronary angiography was performed due to the patient's increasing complaints. Coronary angiography revealed normal coronary arteries, and a fistula between the RCA and the bronchial vein (*Figures 1–3*). Due to the patient's symptoms recurring, we thought that the fistula may have reopened and be causing myocardial ischemia (*Figure 4*). Closure of the fistula using catheter-based methods (transcatheter coil embolization) was planned. The patient was discharged under medical treatment, and was given an appointment for the procedure.

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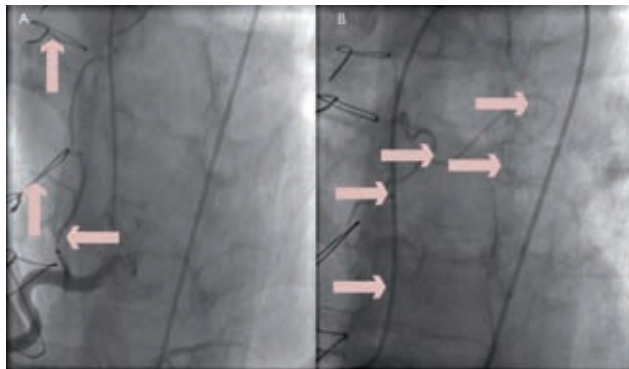
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Figure 1: Non-atherosclerotic Coronaries



A: Normal left main coronary, circumflex, left anterior descending artery; B: Normal right coronary artery. No atherosclerosis narrowing or lesions can be seen.

Figure 3: The Steel Wire Link Suture from the Previous Coronary Fistula Ligation Operation and Course of the Fistula

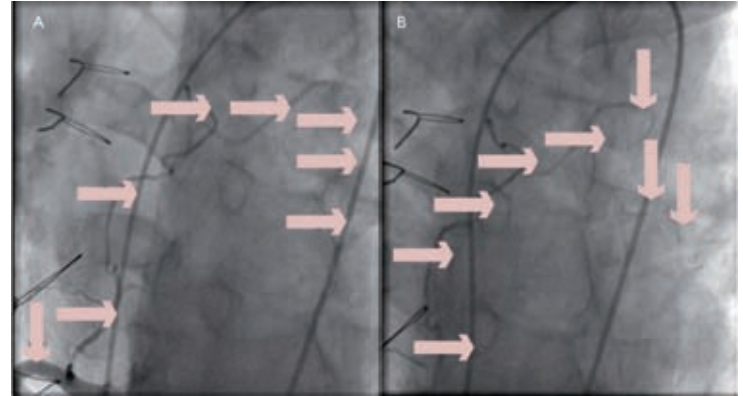


A: The steel wire link suture from the previous coronary fistula ligation operation (vertical) and the location of the coronary fistula ligation showing late-term recanalization (horizontal arrow); B: The navigation of the fistula between the right coronary artery and the bronchial vein can be seen here (horizontal arrows).

Discussion

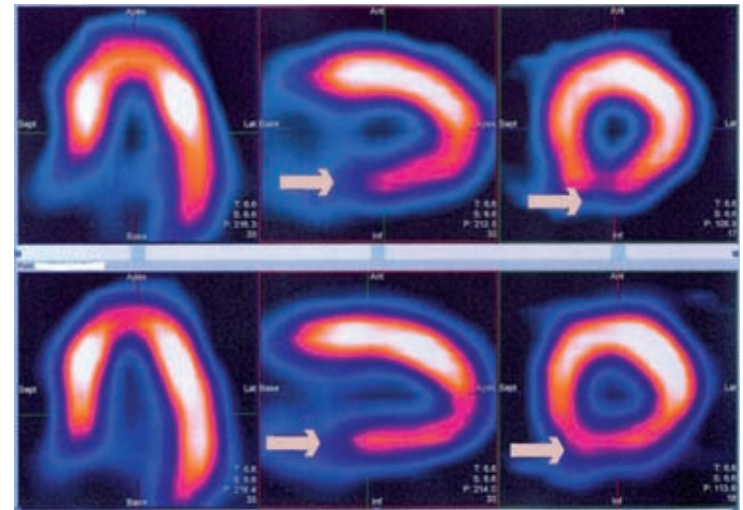
A CAF is the establishment of a direct link between the coronary arteries and the cardiac chambers, mostly to the right ventricle, then the right atrium, the pulmonary artery, and the coronary sinus, respectively. CAFs are found in approximately 0.1 % of patients who undergo coronary angiography.³ CAFs originate from the right coronary artery and its branches in 55 % of cases, the left coronary artery in 35 % of cases, and the remainder originate from both coronary arteries. The link between the coronary arteries and the cardiac chambers originates from embryonic developmental period. Fistulas mostly open towards the right ventricle, the right atrium or the coronary sinus. Fistulas from the coronary artery to the pulmonary artery are generally identified coincidentally during coronary angiography.⁴ The shunts in CAFs are usually small and do

Figure 2: Onset and Course of the Fistula Between the Coronary Artery and Bronchial Vein



A: The fistula starting from the proximal of the right coronary artery (vertical arrow) and ending in the bronchial vein (horizontal arrow) can be observed; B: After the coronary fistula navigation (horizontal arrow), the opening to the bronchial vein (vertical arrow) can be seen.

Figure 4: Myocardial Perfusion Scintigraphy



A myocardial perfusion defect can be seen in the inferior left ventricular segment stress image (horizontal arrows) and this perfusion defect is corrected on the rest images. The myocardial perfusion defect is not seen in the other segments (apex, septum, lateral, or anterior segments).

not cause myocardial ischemia, and so patients may be asymptomatic. However, if there is a big left to right shunt, many complications such as congestive heart failure, pulmonary hypertension, bacterial endocarditis, and fistula thrombosis can occur. Also big shunts can cause myocardial ischemia due to a decrease in coronary blood flow (coronary steal syndrome).⁵ In coronary steal syndrome, shunt flow changes from ischemic to normal region. Consequently myocardial blood flow reduces and ischemia occurs.⁶

Most pediatric patients are asymptomatic, and so a CAF should be considered as a differential diagnosis when a heart murmur is heard during examination. In adult patients, a CAF may cause symptoms such as chest pain on exertion (angina), or shortness of breath. In one study, 70 % of patients with identified CAFs had angina pectoris (chest pain) and shortness of breath, and 75 % of patients had non-specific electrocardiography variations. Myocardial ischemia was determined in 29 % and 50 % of patients during exercise stress testing, and thallium stress scintigraphy, respectively. Atherosclerosis can occur in the coronary artery where the fistula is located. When this is the case, heart attacks have been reported in 15 %.⁷ The long-term prognosis of asymptomatic or small CAFs is better. In one study, patients who had small, coincidentally identified CAFs were followed up for nine years. Echocardiography was normal and 23 % of small CAFs closed by themselves during the follow-up period. Untreated big CAFs may cause early coronary artery disease. The most important factors that affect spontaneous closure of CAFs is the size and flow rate. If closure of asymptomatic fistulas is considered, the risks and benefits of the procedure should be discussed with the patient.

For the patient discussed in this case report, the reason for closure of the fistula was anginal pain that was refractory to medical treatment. Fistulas can be closed with embolytic material (transcatheter coil embolization) or surgically. In

one study, the long-term (17-year) prognosis was investigated in children with CAFs treated surgically. Many complications may occur as a result of surgery, such as recanalization, dilatation in the coronary artery ostium and other segments, thrombus formation, calcification, and myocardial infarction.⁸ The rate of fistula recanalization due to surgery was reported to be up to 9 %. Surgery and direct epicardial or endocardial ligations were traditionally the main therapeutic methods for closure of CAFs. Surgical correction is safe and effective. However, transcatheter occlusion of CAFs in pediatric patients has been recommended as the preferred therapeutic option. Compared with surgery, transcatheter closure is less invasive, less prone to complications, and results in a shorter hospital stay. The main indications for closure are clinical symptoms, especially heart failure, and myocardial ischemia. The success rate of transcatheter closure of coronary artery fistulas was reported as 90 %. Peri-procedural death was not reported.⁹

In the literature, a case of fistulization from the coronary to the bronchial artery has been reported.¹⁰ However, the case discussed here has not been reported in the literature so far.

In conclusion, CAFs may be symptomatic and cause myocardial ischemia, especially when they are large. Recanalization of the fistulas may occur after surgical closure or the transcatheter approach, even later on. This should always be kept in mind. ■

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