

Case Report

Therapeutic transcatheter embolization of bilateral coronary-to-pulmonary fistula with symptomatic cardiac dysfunction in two adults

Mingyi Shang^{1*}, Jinhong Wang^{2*}, Bo Zhang², Jun Ma¹, Jinfa Jiang³

¹Department of Interventional Radiology, Shanghai Tongren Hospital, Shanghai 200336, China; ²Department of Medical Imaging, Tongji Hospital, School of Medicine, Tongji University, Shanghai, China; ³Department of Cardiology, Tongji Hospital, School of Medicine, Tongji University, Shanghai, China. *Equal contributors.

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Abstract: Bilateral coronary-to-pulmonary artery fistulas are extremely rare in congenital malformation among coronary artery fistulas. We reported two cases with symptomatic cardiac dysfunction were treated successfully by therapeutic transcatheter embolization. Two patients with bilateral coronary-to-pulmonary artery fistulas were admitted to our hospital. Patient 1 showed the reduced left ventricular diastolic function. Patient 2 showed a reduced left ventricular systolic function with moderate pulmonary hypertension and severe tricuspid regurgitation. ECG showed a pacing ECG with the complete right bundle branch block, anterior wall myocardial infarction, anterior wall and inferior wall cardiac ischemia as well as enlarged cardiac silhouette in its Chest X-ray. Coronary angiography revealed bilateral tortuous coronary fistulas originated from the left anterior descending artery and the right coronary artery in both patients, and drained into the main pulmonary artery. The two patients were treated successfully by therapeutic transcatheter embolization. We considered that selected transcatheter closure of coronary artery fistulas is a minimal invasive and feasible treatment in the presence of heart failure. Clinical and cardiac assessment should be done before transcatheter closure.

Keywords: Coronary artery fistula, angiography, coil embolization

Introduction

Coronary artery fistulas (CAF) are rare congenital or acquired disorders with single or multiple abnormal passages between coronary artery and a cardiac chamber or vessels. Bilateral fistulas, originating from left and right coronary arteries, are extremely rare (only 5% of CAF). These fistulas are orientated to communicate with the pulmonary artery [1]. Their clinical significance usually depends on the risk of complications such as arrhythmias, infectious endocarditis, myocardial ischemia, sudden death, congestive heart failure and so on. At present, open surgical repair is considered as the mainstream of treatment in the presence of symptoms, aneurysm, heart failure and preventive complication [2, 3]. However, damage control treatment has been slow to be accepted by surgeons around the world as the increasing advances in transcatheter technique in recent

decades. We present our experience to treat two adult patients with reduced heart function secondary to the bilateral coronary-to-pulmonary artery fistulas by using the minimal invasive procedure, namely therapeutic transcatheter embolization of CAF.

Case presentation

Case 1

A 57-year-old female complained of episodes of chest pain, dyspnea, tachycardia and heart murmur. Her physical examination was unremarkable as well as chest X-ray. Electrocardiography showed a normal sinus rhythm. Cardiac enzymes were in normal range. Cardiac catheterization exam showed a higher pulmonary capillary pressure and pulmonary arterial hypertension. Echocardiogram revealed the reduced left ventricular diastolic function.

Embolization for coronary fistula

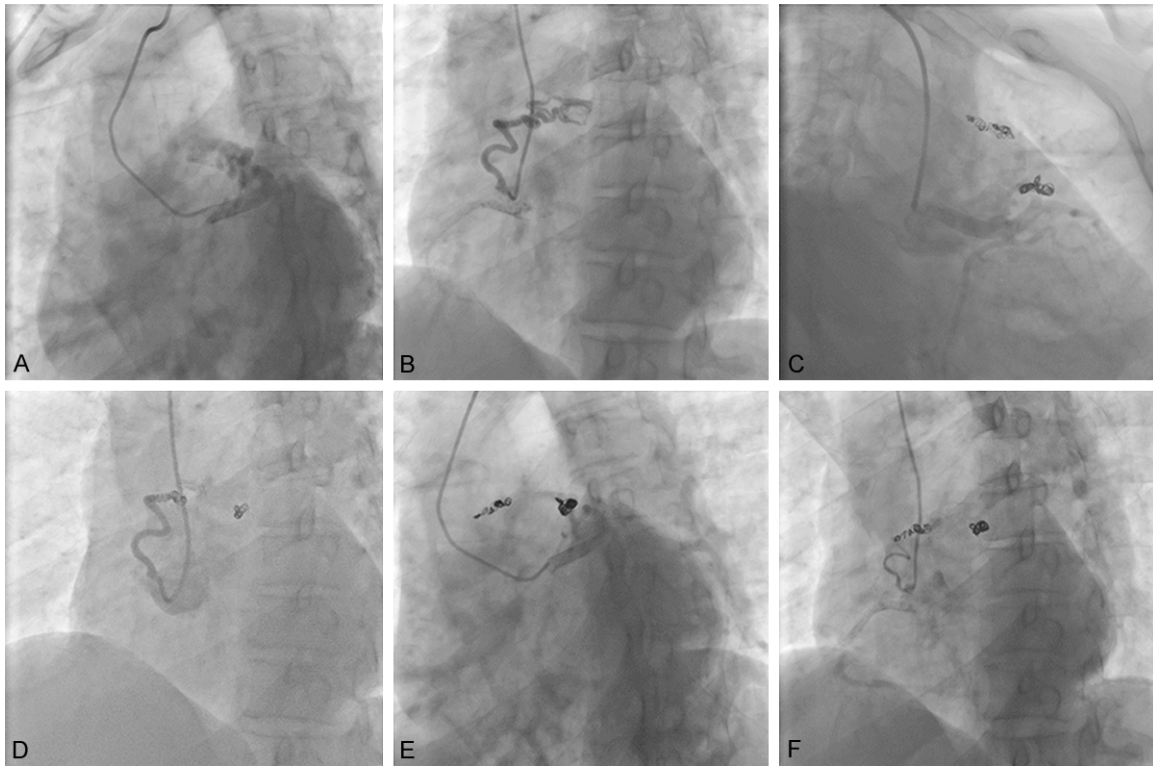


Figure 1. A. Left coronary angiogram showed a coronary artery fistula originating from the proximal left descending coronary artery draining into the main pulmonary artery. B. Right coronary angiogram showed a coronary artery fistula originating from the right coronary artery terminating in the main pulmonary artery. C. Left coronary angiogram showed that packing coils resulted in occlusion of the fistula, but the distal site had a little blood flow. D. Right coronary angiogram showed that packing coils resulted in complete occlusion of the fistula. E. At 5-month follow up, Left coronary angiogram showed that the fistula was embolized by the coils mostly, but the distal site had a little blood flow, which was the same as before. F. Right coronary angiogram showed the fistula was completely occluded by the coils.

Coronary angiography demonstrated two fistulas between bilateral coronary artery and pulmonary artery (**Figure 1A, 1B**). Anomalous vessels originated from the both proximal bulky left anterior descending artery and the right coronary artery respectively, and drained into the main pulmonary artery. After obtaining the patient's consent, therapeutic transcatheter embolization was performed. A right radial approach with 6F Judkins guiding catheter was used to open the left coronary artery. A 0.014" guide wire was advanced through the guiding catheter to the distal segment of the fistula, and the micro catheter was sent through the guide wire. After confirming the micro catheter's position by angiography, coil embolization was then performed by releasing the coil named MWCE-18S-6/2-TORNADO. The coil passed through micro catheter to the sinus. According to the blood flow conditions, coils 6/2, 6/2, 4/2, 4/2 were successively added to

the same position. Imaging the patient, some contrast agents was still in the fistula. Coils were fixed well within the blood vessels. The distal yet had a little blood flow (**Figure 1C**). Then we dealt with the fistula from the right coronary artery to close the right fistula. The micro catheter was placed through the guide wire. After confirming the position by the angiography, coil embolization was performed using the coil named MWCE-18S-6/2-TORNADO. According to the blood flow conditions, coil 4/2, 4/2, 4/2, 3/2 were added successively. Imaging the patient, there were some contrast agents in the vascular of the fistula. 10 minutes later after confirming that the coils were implanted in a suitable position and were fixed well within the blood vessels, selective coronary angiography was performed. The angiography showed a complete occlusion of the fistula (**Figure 1D**) after the embolization. There were no signs of myocardial damage. ECG showed sinus rhythm

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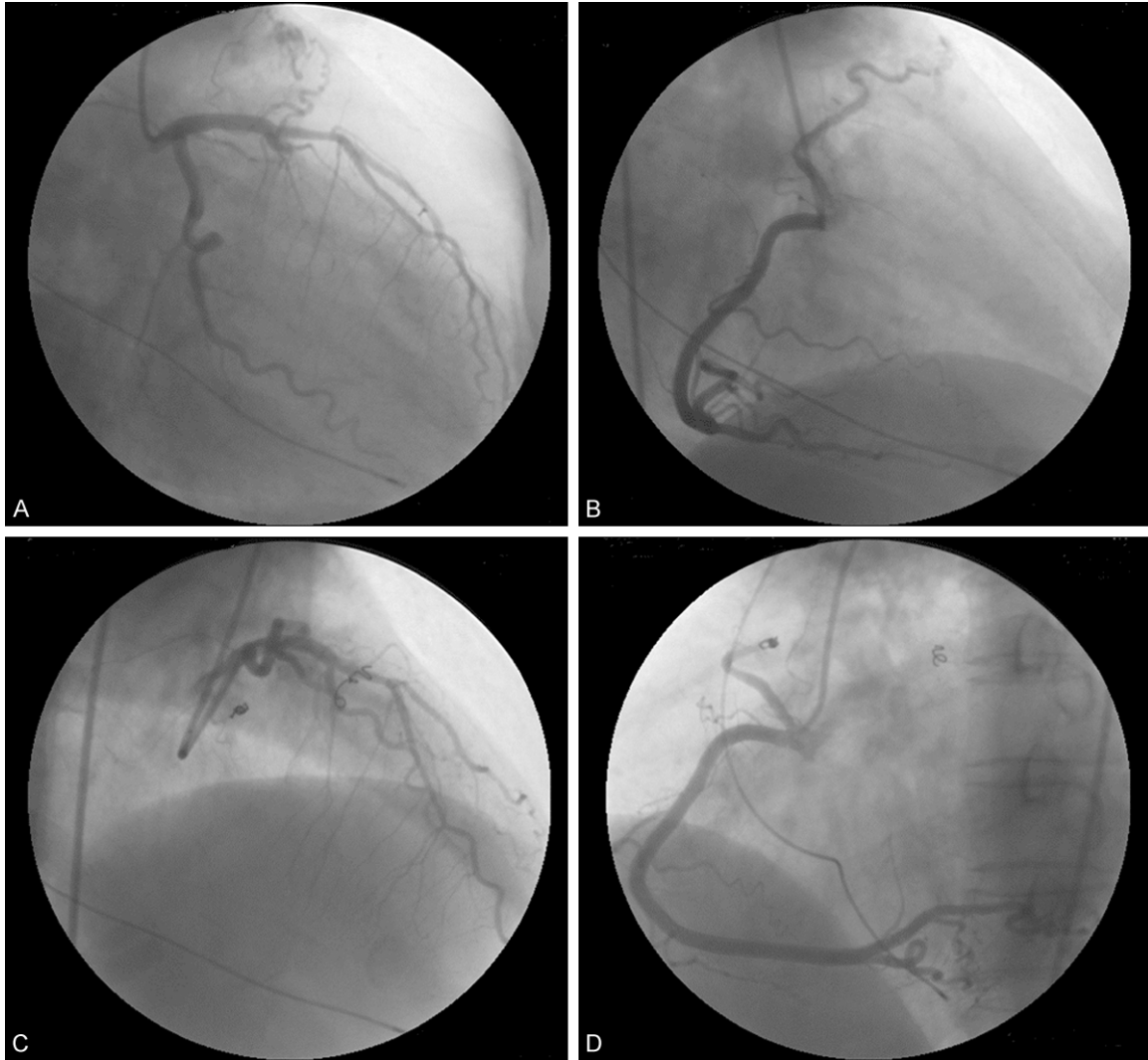


Figure 2. A. Left coronary angiogram showed a fistula between the proximal left anterior coronary artery and the pulmonary artery trunk. B. Right coronary angiogram demonstrated coronary artery fistula originating from the right coronary artery and terminating in the main pulmonary artery. C. After coils embolization, left coronary angiogram showed a complete occlusion of the fistula. D. Right coronary angiogram showed that packing coils resulted in complete occlusion of the fistula.

with T wave anomaly and insignificant ST segment. Echocardiography revealed an enlarged left atrium with 9 mm in diameter. The patient's left ventricular systolic function showed normal, left ventricular diastolic function became lower and left ventricular ejection fraction was 70%. The patient's dyspnea has been rapidly alleviated, pulmonary pressures became normal. Seven days later after the transcatheter coil occlusion, patient in good health was discharged from the hospital and was prescribed to take aspirin at least 6 months to 9 months. To further clarify the fistulas condition, the patient was asked to have a selective review after 5-month.

At 5-month follow up, the patient was free of chest pain and dyspnea. Chest X-ray, ECG and cardiac enzymes were within normal limits. A selective coronary angiography showed that the coil embolization was in suitable condition and showed a bulky left anterior descending branch trunk. There was still a little blood flow in the fistula (**Figure 1E**) after the coil embolization. Right coronary artery angiography after the embolization demonstrated complete occlusion of the fistula (**Figure 1F**). Left ventricular angiography with pigtail catheter showed normal left ventricular systolic function and the left ventricle ejection fraction of 61%. Physical examinations demonstrated the patient was in

good conditions after the interventional treatment.

Case 2

A 61-year-old diabetic female with hypertension was admitted to our emergency room because of weakness, dizziness, atypical chest pain, exertional angina and dyspnea. She had a history of permanent pacemaker implantation and renal dysfunction. Cardiac enzymes were within normal limits. Chest X-ray showed a significantly enlarged cardiac silhouette. 12-lead electrocardiogram (ECG) revealed a paced rhythm, atrial fibrillation, complete right bundle branch block, anterior wall myocardial infarction, anterior wall and inferior wall cardiac ischemia. Dynamic electrocardiogram showed atrial flutter, cardiac ischemia and occasional premature ventricular contractions. A reduced left ventricular systolic function with moderate pulmonary hypertension and severe tricuspid regurgitation are demonstrated. Coronary angiography revealed two fistulas with one fistula originating from the proximal left anterior descending artery (LAD) (**Figure 2A**) and another one from the right coronary artery (**Figure 2B**) and draining into main pulmonary artery. Therapeutic transcatheter embolization was undertaken because of her dysfunctional heart and kidney. In this procedure, a 6FJL4.0 and a JR4.0 guiding catheter was used. The left coronary artery fistula was first totally occluded using 2 coils (**Figure 2C**). The right coronary artery fistula was occluded completely by using 3 coils (**Figure 2D**). The other procedure was similar to the patient 1. We observed the patient's clinical symptoms rapidly and significantly improved and her pulmonary artery pressures were back to normal. 2-D Echocardiography showed normal volume in left atrial and ventricles. The patient had been following up until a lung cancer was diagnosed recently.

Discussion

Coronary-pulmonary artery fistula as an anomalous origin of the coronary artery from the pulmonary artery, is commonly considered under the grouping of coronary artery fistula. Bilateral coronary-pulmonary artery fistulas are distinctly rare. In childhood, most patients with coronary artery fistulas remain asymptomatic. Spontaneous closure may occur in small fistula. Follow-up observation is necessary for these

patients. About half of coronary artery fistulas develop cardiovascular symptoms as patients age [4]. Based on the recent published articles [1-5], most of these fistulas were treated by various surgical procedures such as fistula closure with or without cardiopulmonary bypass and closure under pulmonary arteriotomy. Usually, operations are indicated in the presence of large fistulas, myocardial ischemia, aneurysmal formations or heart failure due to shunt flow. Progressively enlarging fistulas, the presence of symptoms, aneurysm, heart failure and preventive complication warrant operative repair.

Currently, more and more about transcatheter closure of coronary artery fistulas were reported [1, 6]. With the current development of interventional devices and increasing techniques for transcatheter closure, similar results to surgery have been achieved with minimal invasive and a shorter recovery time [6]. Furthermore, the transcatheter closure is a preferred technique nowadays, particularly in the absence of multiple fistulas and large fistula branches [7]. The advantages of this technique include minimal invasion, real time observation and management, dedicated manipulation, high closure rate, and low morbidity and mortality rates. An experienced operator and interventional specialist with expertise in coronary arteriography, embolization techniques and complete preoperative evaluation can warrant a successful transcatheter approach. However, patients with large fistula, multiple openings, or significantly aneurysmal dilation may not be indicated for transcatheter closure.

In our series, surgery was deferred in patient 1 and 2 because of moderate to severe cardiac dysfunction and diabetes. In order to carry out an iatrogenic damage control, therapeutic transcatheter embolization instead of surgery was performed after complete evaluation of cardiac and other organ function. We managed to initially build a nest by using oversized coils to reduce the risk of escape of the coil. Then, we selected many small and adequate coils to achieve the completely embolization. Postoperatively, we achieved the goal of treatment of minimal invasion, few fistulas left and rapid symptomatic improvements on the follow-up evaluation.

In conclusion, we considered that selected transcatheter closure of CAF is a minimal inva-

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sive and feasible treatment in the presence of heart failure. Clinical and cardiac assessment should be done before transcatheter closure.

Disclosure of conflict of interest

None.

Address correspondence to: Mingyi Shang, Department of Interventional Radiology, Shanghai Tongren Hospital, 1111 Xianxia Road, Changning District, Shanghai 200336, China. Tel: +86-13816294467; Fax: +86-021-64085875; E-mail: myshang1969@aliyun.com

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