



CASE REPORT

Congenitally severe tortuous circumflex artery fistula draining into the coronary sinus: Transcatheter closure with Guglielmi detachable coils via different delivery system

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Summary Coronary artery fistulae are very rare congenital anomalies which constitute 0.2–0.4% of all congenital heart diseases. The right chambers of the heart are the most frequent communication site of the coronary fistulae and may cause hemodynamic impairment in the coronary circulation. The fistulae arise frequently from right coronary and left anterior descending arteries. Fistulae from left circumflex artery are very rare. We report a case in which transcatheter embolization was performed with 3-mm Guglielmi detachable coils in a young adult with a rare type of congenital fistula originating from the circumflex artery and draining into the coronary sinus.

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Introduction

Coronary artery fistulae are rare congenital heart diseases [1–4]. The most frequent communication sites are right chambers of the heart. Fistulae from

left circumflex artery are very rare. The communication size and location of the coronary fistulae determine the hemodynamic significance. Large coronary artery fistulae may cause congestive heart failure, myocardial infarction, arrhythmias, infectious endocarditis, or rupture. Surgical or transcatheter closure of coronary artery fistulae are treatment approaches to prevent hemodynamic impairment. Transcatheter closure may be performed with many percutaneous interventional embolic techniques [5–7]. Large clinical trials have shown

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the safety and efficacy of Guglielmi detachable coils [8]. We describe a case in which transcatheter embolization was performed with 3-mm Guglielmi detachable coils in a young adult with a rare type of congenital fistula originating from the circumflex artery and draining into the coronary sinus.

Case report

The patient, a 35-year-old male was referred for chest pain. He complained of chest pain during exercise for one year. On physical examination his blood pressure was 110/70 mmHg, and pulse 80 beats/min. There was no other pathologic sign. Posterior to anterior chest X-ray demonstrated normal pulmonary conus. Sinus rhythm and 1 mm ST elevation in D II, aVF leads were noted on electrocardiography. Other laboratory findings were normal.

Transthoracic echocardiography showed mildly dilated right heart chambers and there was no other pathologic finding. Myocardial perfusion scintigraphy demonstrated ischemia in anterolateral territory. At catheterization, the femoral artery and vein were cannulated. Selective coronary angiography of the left coronary artery revealed the severe tortuous fistula arising from the distal portion of circumflex artery and draining into the coronary sinus (Fig. 1). Cardiac catheterization was performed to assess the value of left to right shunting. Pulmonary

artery pressure was 25 mmHg and arterial pressure was 110/70 mmHg. O₂ saturation was 75.5–74.7% in superior vena cava, 73.8–79.3–81.5% in right atrium and right ventricle, 78% in vena cava inferior, 78.6% in pulmonary artery, and 97% in femoral artery. There was mild oximetric step-up in the right atrium. The pulmonary to systemic blood flow ratio (QP/QS) was calculated and found to be 1.3. The patient complained of exertional dyspnea and had dilated right side of the heart. Therefore, we decided to close the coronary fistula with a transcatheter approach.

A 6 Fr guiding catheter was used to cannulate the left coronary system. Thereafter, a 2.0 Fr infusion microcatheter (Excelsior 1018, Boston Scientific, Natick, MA, USA) was introduced through the guiding catheter over a 0.012 in. hydrophilic guidewire with a 90° tip (Radiofocus, Guidewire M, Terumo, Tokyo, Japan) until it was selectively placed within the fistula. Embolization was accomplished with four 3-mm Guglielmi detachable coils (Boston Scientific/Target, Cork, Ireland). Multiple coil placement was necessary for closing of the severe, tortuous, high-flow fistula. After occlusion of the fistula was confirmed with angiography via guiding catheter, the microcatheter was removed. Repeat selective left coronary angiography after removing the microcatheter, also revealed complete occlusion of the fistula (Fig. 2). The patient was discharged on the second day of the procedure

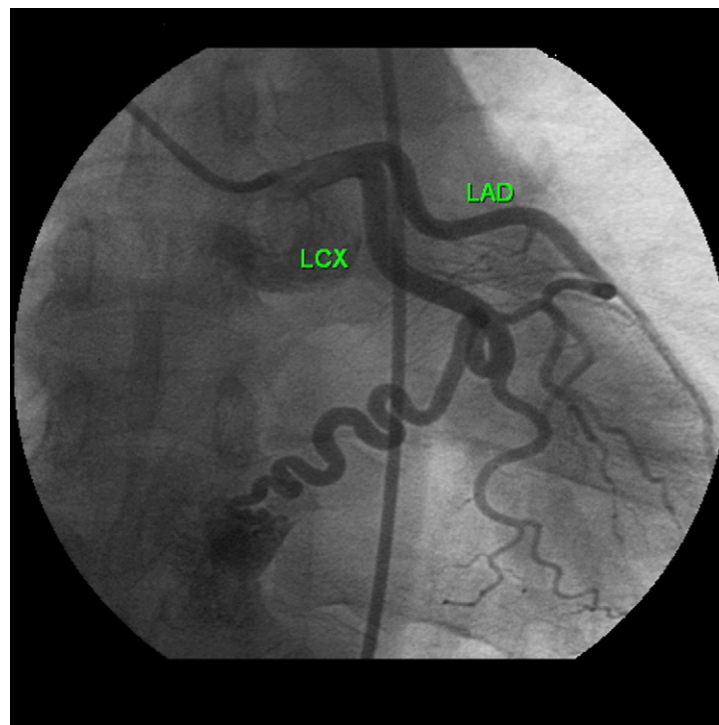


Figure 1 Severe tortuous fistula arising from distal circumflex artery.



Figure 2 Coronary angiography showing complete occlusion of the fistula.

without complications. After one month, in selective coronary angiography, there was a blood flow due to partially occluded fistula (Figs. 3 and 4). The patient was asymptomatic and the blood flow from the fistula had low velocity with QP/QS 1.1, therefore medical treatment was preferred.

Discussion

Coronary artery fistulae are rare cardiac anomalies and the first case was described in 1865 [9]. Closure of coronary fistulae during childhood is recommended because of symptoms and



Figure 3 After one month partially occluded fistula in RAO view.

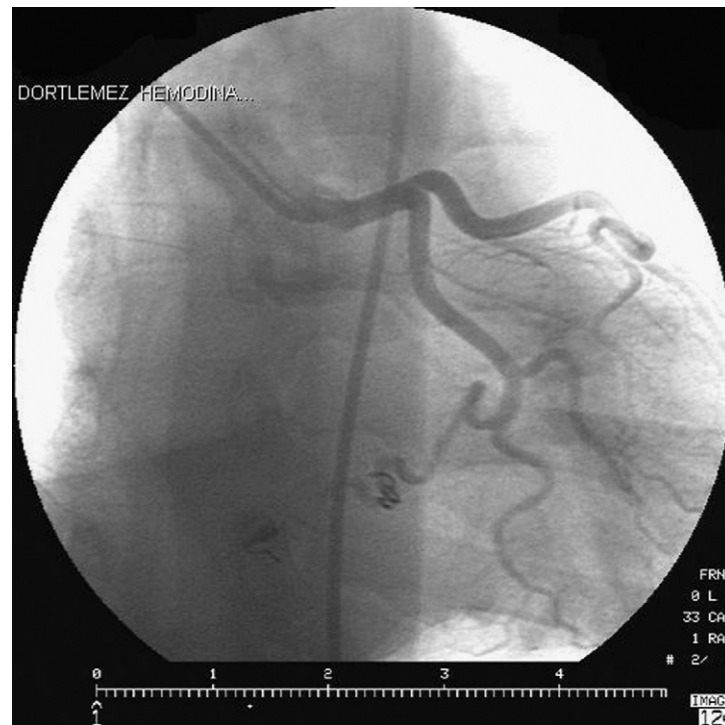


Figure 4 After one month partially occluded fistula in caudal view.

complications due to hemodynamic impairment. Treatment approaches are surgical repair or catheter based closure in congenital coronary fistulae. Transcatheter closure of coronary fistulae with many percutaneous interventional embolic occlusion techniques with various materials including Gianturco coils, polyester-covered stainless steel coils, detachable balloon, double umbrella devices, polyvinyl alcohol particles, and glue have been reported [5–7].

The Guglielmi detachable coil system consists of a soft platinum coil soldered to a stainless steel delivery wire. When the coil is properly positioned, a 1 mA current is applied to the delivery wire. The electric current dissolves the stainless steel delivery wire proximal to the platinum coil by means of electrolysis. Mechanical detachable coils were used in patients with high-flow velocity coronary fistulae and had an important role in avoiding coil migration and safe coil embolization [10,11]. Electrically detachable coils are particularly safe with minimal risk of migration due to high-flow velocity [12,13]. In this case, transcatheter closure was performed with Guglielmi detachable coils. Large clinical trials showed the safety and efficacy of Guglielmi detachable coils in endovascular treatment [8]. In the present case, we preferred the Guglielmi detachable coils in order to prevent unintended embolization of the parent circumflex artery as this

system allows the detachment of coils in the exact position and withdraw it if the result is unsatisfactory. Particularly in high-flow cases, angiographic follow-up is necessary for evaluation of reopening of coronary fistulae. This issue must be kept in mind for long-term management of patients with large coronary fistulae.

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