How to treat the combination of coronary artery fistula and occluded coronary artery
A case report
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Abstract
Rationale: Coronary artery fistulae (CAF) are uncommon and are reported in 0.25% of patients undergoing routine coronary angiography. The combination of severe coronary artery disease and a CAF was rare.

Patient concerns: A 76-year-old man presented unstable angina.

Diagnoses: Coronary angiography showed a subtotal occlusion lesion and a CAF at left anterior descending artery.

Interventions: The combination of transcatheter coil embolization and a drug-eluting stent was used for this problem.

Outcomes: The patient was symptom-free and had regularly followed up as an outpatient for three years.

Lessons: The combination of a CAF and atherosclerotic change in the same site of the coronary artery is unique. It is feasible to solve the combination of CAF and severe coronary atherosclerotic lesion by transcatheter coil embolization and stent implantation. A big case series study is necessary to evaluate the safety and efficacy of the technique of transcatheter coil embolization and stent implantation in the patients with severe coronary atherosclerotic lesion and a CAF.

Abbreviations: CAF = coronary artery fistulae, CAG = coronary angiography, DES = drug-eluting stent, IVUS = intravascular ultrasound, LAD = left anterior descending artery, LCX = left circumflex artery, NSTEMI = non-ST-segment myocardial infarction.

Keywords: coronary arterial fistula, drug-eluting stents, severe coronary artery disease, subtotal occlusion, transcatheter coil embolization

1. Introduction
Coronary artery fistulae (CAF) were first described by Krause in 1865.[1] According to Ogden’s classification of congenital anomalies of the coronary arteries, CAF are an anomaly of termination, representing abnormal communication between the coronary arteries and the cardiac chambers (coronary-cameral fistulae) or low-pressure veins (coronary arteriovenous malformations).[2] In the literature review, the combination of severe coronary artery disease and a CAF was reported rarely. In our case report, we reported a 76-year-old man, who presented unstable angina, received coronary angiography and showed a subtotal occlusion lesion combined a CAF at left anterior descending artery. Transcatheter coil embolization was performed first for CAF, and a drug-eluting stent was deployed for coronary artery lesion. It is feasible to solve the combination of CAF and severe coronary atherosclerotic lesion by transcatheter coil embolization and stent implantation.

2. Case description
A 76-year-old man presented with intermittent chest discomfort that worsened with exercise for the past 2 years. He had a medical history of type 2 diabetes mellitus and hypertension for several years. The transthoracic echocardiography showed adequate left ventricular performance, but some abnormal flow at the pulmonary artery root. The coronary angiography (CAG) showed 60% stenosis at the midportion of the right coronary artery, which supplied the distal left anterior descending artery (LAD) (Fig. 1A). The proximal LAD had one coronary artery to pulmonary arterial fistula and a subtotal occlusion at its proximal part, as well as 60% to 70% stenosis at the proximal left circumflex artery (LCX) (Fig. 1B and C). A 0.014 Fr. Fielder FC wire (Asahi, Intecc) with a microcatheter (Renegade, Boston) was passed through the tight lesion carefully. After ballooning, the CAF was still visible despite the improvement in the antegrade flow of the coronary artery...
The intravascular ultrasound (IVUS) (Volcano, Boston, San Diego) showed multiple densely calcified plaque and the origin of the fistula (Fig. 1E).

Initially, we chose a cover stent for or a drug-eluting stent (DES) for this rare combination. When considering of the poor clinical outcome of cover stent implantation and the lesions with severe calcification, we chose the strategy of DES implantation. However, we were afraid of that the CAF could not be sealed after a DES use. Therefore, we decided the strategy of the combination of transcatheter coil embolization and DES use.

Then, 3 coils (2: 3.0 × 2.5 mm; 1: 4.0 × 4.0 mm; VortX 18, Boston) were used to obstruct the CAF (Fig. 2A) via the microcatheter. One Everolimus-eluting stent (2.5 × 38 mm, Xience Prime, Abbott) was placed at the proximal to mid-LAD (Fig. 2B). After postdilatation and stenting at the mid-LAD, the final CAG showed TIMI 3 flow of the left coronary artery, and occlusion of the CAF (Fig. 2C and D). The patient was symptom free and had regularly followed-up as an outpatient for three years.

3. Discussion

Congenital anomalies of the coronary arteries occur in 0.2% to 1.2% of the general population. Most CAF drain into the right heart chamber or into the pulmonary artery and arise from the right coronary artery (50%); fewer than half arise from the circumflex artery, and 5% arise from both coronary arteries. Most of these anomalies do not cause myocardial ischemia; often, they are found incidentally during angiographic evaluation for other cardiac diseases. Treatment of CAF is indicated for symptomatic patients and for those asymptomatic patients with significant shunt or large fistulas that create a risk for future complications, such as infective endocarditis, pulmonary hypertension or heart failure.

Transcatheter closure approaches have emerged as a less invasive and effective strategy. The advantages of the transcatheter approach include less morbidity, lower cost, shorter recovery time, and avoidance of thoracotomy and cardiopulmonary bypass. Several devices including coils, detachable balloons, covered stents, vascular plugs, and atrial septal defect devices had been used for the closure of CAF based on their size. Only 2 case reports presented the combination of severe coronary disease and CAF. One case presented non-ST-segment myocardial infarction (NSTEMI) and diagnosed a CAF from left main, which also presented severe stenosis, and treated by cardiovascular surgeon. Another case also presented NSTEMI and diagnosed a large fistula and adjacent atherosclerotic plaque with stenosis at LAD, and treated with a covered stent.

In our case, the patient had multiple risk factors for coronary artery disease and densely calcified plaque at the tight lesion by IVUS image. The clinical outcomes of covered stent for coronary perforation are not very well if the patient had multiple risk factors or severe coronary artery disease. Therefore, the strategy of combination of coils and DES were used for this rare condition, because of the high probability of restenosis.

4. Conclusion

The combination of a CAF and atherosclerotic change in the same site of the coronary artery is unique. It is feasible to solve the combination of CAF and severe coronary atherosclerotic lesion by transcatheter coil embolization and stent implantation. A big case series study is necessary to evaluate the safety and efficacy of the technique of transcatheter coil embolization and stent implantation in the patients with severe coronary atherosclerotic lesion and a CAF.
Author contributions

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Figure 2. Coronary angiography and percutaneous transcatheter intervention. (A) CAG: Coils used to obstruct the coronary arterial fistula (white arrow). (B) CAG: A drug-eluting stent (2.5 × 38 mm, Xience Prime, Abbott) was deployed at the proximal to midportion of the LAD (black arrow), and deployed coils (white arrows). (C and D) CAG: The left coronary artery showing fair flow, and occluded flow of the coronary arterial fistula. CAG = coronary angiography.