

Coronary Artery Fistula with Giant Aneurysm and Coronary Stenosis Treated by Transcatheter Embolization and Stent

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Coronary artery fistula (CAF) with giant aneurysm and accompanied by coronary artery stenosis is a very rare disease. Herein, we report a case of a 76-year-old woman having a complex coronary-to-pulmonary artery fistula associated with a giant aneurysm and accompanied by coronary artery stenosis. The patient was successfully treated using transcatheter coil embolization and coronary stent implantation. Eight years later, we performed a follow-up coronary angiogram, which revealed the CAF and the aneurysm were completely occluded and previous stent patency. (**Korean Circ J 2015;45(3):245-247**)

KEY WORDS: Arteriovenous fistula; Coronary aneurysm; Embolization, therapeutic; Therapeutics; Stents.

Introduction

Coronary artery fistula (CAF) is defined as a communication between a coronary artery and a great vessel or a cardiac chamber. CAF is a rare anomaly with an incidence ranging from 0.1–0.2% in patients who undergo coronary artery angiography.¹⁾²⁾ Symptoms and signs are dependent on the size of the fistulous connection. The clinical symptoms of CAF include heart failure, infective endocarditis, angina pectoris, fatigue, and respiratory infection.

Coronary arterial aneurysm (CAA) is defined as a coronary dilatation that exceeds the diameter of the adjacent normal arterial segment or the patient's largest coronary artery by 1.5 times. CAAs are uncommon lesions, noted in 0.2% to 4.9% of patients undergoing coronary angiography.³⁾⁴⁾ "Giant" CAAs, which are

defined as having a diameter of 20 mm or greater, are rarer, with a reported prevalence of 0.02%.⁵⁾ Several cases have reported a spontaneous rupture of a giant aneurysm.⁶⁾

We report a case of 76-year-old woman who developed a CAF with the formation of a giant aneurysm and accompanying stenotic lesions, which was successfully treated by transcatheter coil embolization and implantation of a coronary stent. The long-term outcome appears satisfactory.

Case

A 76-year-old woman with past medical history of hypertension was referred to our hospital due to a new onset of chest pain at rest radiating to her left arm, lasting for more than 20 minutes in March 2006. She had no illness comparable to Kawasaki disease. On arrival, her vital signs were stable with a body temperature of 36.8°C, blood pressure of 140/90 mm Hg, a pulse rate of 72 beats/minute, and a respiratory rate of 20 breaths/minute. The physical examination showed a continuous Levine grade 3/6 heart murmur heard at the second left sterna area. An electrocardiogram showed a normal sinus rhythm with left ventricle hypertrophy by voltage and T-wave inversion in leads V 2–5. Her chest X-ray demonstrated mild cardiomegaly. Her echocardiography showed preserved left ventricular systolic function (ejection fraction=54.3%) with mild left ventricular hypertrophy. Cardiac enzymes were elevated; creatine kinase-MB was 5.85 ng/mL (0–4.87 ng/mL) and troponin I was 3.42 ng/mL (0–0.05 ng/mL).

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Therefore, she was diagnosed with a non-ST-segment elevation myocardial infarction and an early coronary angiogram was performed. It revealed a giant, calcified thrombus, aneurysm originating from a branch of the left anterior descending coronary artery (LAD), which had a fistula connected to the pulmonary artery and severe (90% diameter) stenosis in the mid-LAD segment (Fig. 1). Five days later, we performed a transcatheter intervention. At first, a 7 Fr Judkins left-type coronary guiding catheter (Brite Tip®, Cordis, Miami, FL, USA) was inserted via the right femoral artery and advanced into the left coronary artery. Then, a 0.014-inch coronary guidewire was inserted into the feeding artery, and a 2.7 Fr microcatheter (Progreat®, Terumo Co., Tokyo, Japan) was successfully advanced into the distal end of the feeding artery. The guidewire was withdrawn and two 0.018-inch

coils (4×2 mm and 3×2 mm, Tornado®, Cook Inc., Bloomington, IN, USA) were placed into the proximal part of the feeding artery. After the coil embolization, a 3.5×23 mm sirolimus-eluting stent (Cypher stent, Cordis, Johnson & Johnson, Miami, FL, USA) and a 4.5×16 mm bare-metal stent (Liberte stent, Boston Scientific, Natick, MA, USA) were implanted in the mid-LAD in an overlapping fashion (Fig. 2). The final coronary angiogram showed good distal flow without residual stenosis and the feeding artery of the aneurysm was successfully closed (Fig. 2B). After the procedures, the patient was free of symptoms. She was discharged without complications and has been regularly followed-up at the outpatient clinic. Eight years later, we performed a follow-up coronary angiogram, which revealed a completely occluded aneurysm and well-maintained patency, despite mild stenosis at the implanted stent (Fig. 2C).

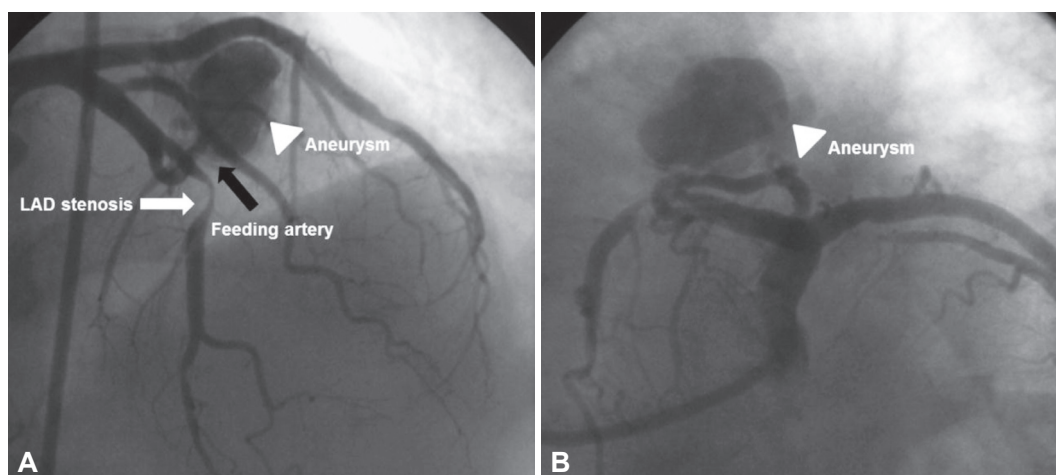


Fig. 1. Left coronary artery angiography demonstrates a giant coronary aneurysm (arrowhead) originating from a branch (black arrow) of the left anterior descending coronary artery (LAD) and severe stenosis in the mid-LAD (white arrow). A: angiography from the anterior-posterior with cranial views. B: angiography from the left-anterior-oblique views with caudal views.

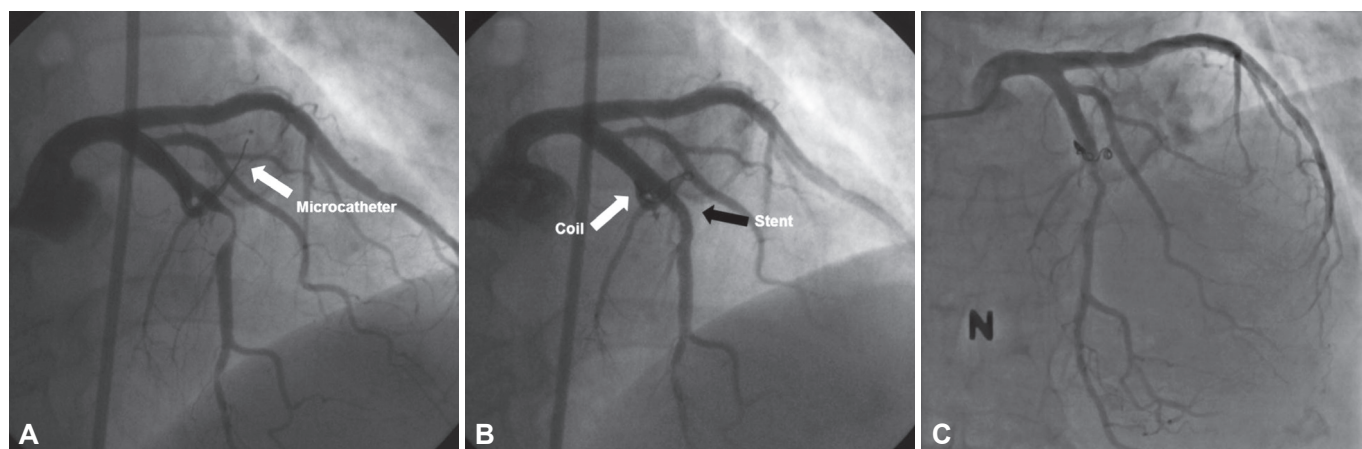


Fig. 2. Percutaneous transcatheter intervention and eight year angiographic follow-up after the procedure. A: a microcatheter is advanced into the distal of the feeding artery (arrow). B: after transcatheter coil embolization (white arrow) and stent implantation (black arrow), a selective left coronary artery angiogram shows successful occlusion of blood flow to the aneurysm cavity by the coils and good distal flow without residual stenosis of left anterior descending coronary artery. C: follow-up coronary angiogram eight years after the procedure showing the aneurysm is completely occluded and well-maintained patency despite mild stenosis at the implanted stent.

Discussion

Coronary artery fistula is accompanied with CAA in approximately 20% of patients.^{7,8)} The mechanism by which an aneurysm forms in the CAF is unclear. It has been suggested that the inflammation, arteriosclerosis, and turbulent blood flow made by vessels' tortuosity and stenosis contribute to aneurysm formation.⁷⁾ For CAF with CAA, surgical ligation or transcatheter intervention may be indicated to prevent spontaneous rupture, dissection, myocardial ischemia, or thromboembolic events.^{6,9)} Surgery ligations were traditionally viewed as the main therapeutic method for the closure of CAF with CAA.^{3,10,11)} Indications for surgery include large CAFs with high flow shunts, multiple communications and terminations, aneurysmal formation, and the need for simultaneous coronary bypass.

Percutaneous transcatheter closure of congenital CAF was first reported in 1983,¹²⁾ and its use is increasing worldwide. This technique can be considered as an alternative to surgery for coronary fistulas because it may decrease the risk of myocardial damage and avoids a thoracotomy and the use of cardio-pulmonary bypass in some cases. The technique is also cost effective, with a short post procedural hospital stay and minimal period of rehabilitation. Therefore, the transcatheter coil closure technique is a valuable alternative to open surgery currently. Indications of transcatheter coil embolization for CAF are absence of multiple fistulae, a single narrow drainage site, absence of large branch vessels, and safe accessibility to the coronary artery supplying the fistula.¹³⁾

Although the transcatheter embolization method is safe and effective for treating CAFs with CAA, complications associated with the procedure, such as embolization of the coil, have been reported, and there is an associated recurrence rate.¹⁰⁾ In spite of an excellent short-term outcome, long-term follow-up is essential due to the possibility of postoperative recanalization, persistent dilation of the coronary artery and ostium, thrombus formation, calcification, and myocardial ischemia. However, only a few cases have reported that catheter intervention with the implantation of a coil for CAF with giant CAA^{9,14)} and the long-term outcome of those cases is still unclear.

Our patient had CAF with a giant CAA and accompanying severe coronary artery disease. Although surgical ligation and coronary artery bypass grafting are still the best method of treatment for these complex diseases, we treated her using transcatheter coil embolization and stent implantation because there was only one feeding vessel, and the aneurysm was proximal to the main coronary vessel and the coronary stenosis was confined to mid-LAD. Since the patient had a high risk of surgical complications due to her advanced age, transcatheter coil embolization and stent implantation appeared to be the better course of action. Coronary angiography, at the our patient's eight year follow-up visit, showed

the CAF and the aneurysm were completely occluded and patent previous stent despite mild stenosis at the implanted stent.

In conclusion, the long-term outcome of a transcatheter coil embolization and stent implantation in CAF with a giant CAA and accompanying coronary stenosis appears satisfactory. Although a case series study is necessary to evaluate the safety and efficacy of the technique, transcatheter coil embolization and stent implantation in these patients is considered to be a promising treatment option.

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