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Case Report

Successful transarterial embolization of coronary artery fistula with ruptured aneurysm: A case report

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We report a case of a coronary artery fistula (CAF) with ruptured aneurysm treated using transarterial embolization (TAE) alone. The ruptured aneurysm caused cardiac tamponade, and it was isolated by embolizing its afferent and efferent vessels using detachable coils. To our knowledge, this is the first case report of successful TAE for a CAF with ruptured aneurysm. We believe that if the patient condition is stable and the CAF is anatomically simple, TAE can be a less-invasive alternative to surgery.

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Introduction

Coronary artery fistula (CAF) is an abnormal vascular shunt from a coronary artery to a cardiac chamber or a great vessel adjacent to the heart. Although CAF is usually congenital and asymptomatic, it can be accompanied by aneurysm formation and in rare cases, aneurysmal rupture, leading to sudden death [1,2]. Some investigators have reported successful surgical treatment for CAF with ruptured aneurysms [3]. Transarterial embolization (TAE) has also been reported in a previous case report; however, surgery was eventually required for the recanalization of the embolization site [4]. Here, we report a case of CAF with a ruptured aneurysm that was successfully treated by TAE alone.

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An 83-year-old woman with a history of hypertension, dyslipidemia, and osteoporosis was admitted to our hospital with unconsciousness, hypotension (68/39 mmHg), and dyspnea (SaO₂ 91% with 6 L/min O₂). Echocardiography revealed cardiac tamponade. Contrast-enhanced computed tomography (CT) showed massive hyperdense pericardial effusion consistent with cardiac tamponade and a CAF arising from the right coronary artery (RCA) to the pulmonary artery (PA) trunk, with an aneurysm measuring 21 mm in diameter. Although no extravasation from the aneurysm was shown, we diagnosed the patient with cardiac tamponade due to the aneurysmal rupture associated with the CAF, because the CT scan revealed no other probable cause of the hemorrhage, such as aortic dissection (Fig. 1A). After pericardiocentesis and hemorrhagic pericardial effusion drainage, the patient's vital signs returned to normal. Multidisciplinary discussion among cardiologists, cardiovascular surgeons, and interventional radiologists led to a consensus to treat the patient using TAE with coils, because the CAF was anatomically simple with a single feeder and a single drainer, and the treatment modality was expected to be less invasive and more prompt than



Fig. 1 – (A) Contrast-enhanced computed tomography (CT) shows massive hemorrhagic pericardial effusion and a fistula from the right coronary artery (RCA) to the pulmonary artery (PA) with an aneurysm (arrow). (B) RCA angiography shows the saccular aneurysm (arrow) with an abnormal afferent vessel (white arrowheads) arising from the proximal portion of the RCA and a tortuous efferent vessel (black arrowheads) draining into the PA trunk. (C) After embolization of the afferent and efferent vessels, RCA angiography shows the disappearance of the aneurysm and coronary artery fistula. (D) After treatment, contrast-enhanced CT shows the disappearance of the enhancement in the aneurysm (arrow).

surgery. A written informed consent was obtained from the patient.

The following interventional procedures were performed through a collaboration between the cardiologists and interventional radiologists. A 6-Fr guiding catheter (Mach 1; Boston Scientific, Natick, MA) was inserted into the RCA via the right femoral artery. Selective angiography showed the saccular aneurysm with an abnormal afferent vessel arising from the proximal portion of the RCA and a tortuous efferent vessel draining into the PA trunk (Fig. 1B). Because the guiding catheter was unstable in the RCA, a 4.2-Fr catheter (Fubuki; Asahi Intecc, Tokyo, Japan) was coaxially inserted into the afferent vessel and advanced close to the aneurysm. Selective angiography showed no myocardium-feeding arteries arising from the afferent artery. Subsequently, a microcatheter (Excelsior 1018, Boston Scientific) was coaxially advanced into the efferent vessel through the aneurysm. The efferent vessel was embolized with 5 detachable coils, 3 mm in diameter and up to 10 cm in length (Target; Stryker, Fremont, CA). Consequently, the afferent vessel was also embolized with 3 detachable coils of 4 mm diameter and 15 cm length (Target; Stryker). The afferent vessel angiography showed the successful isolation of the aneurysm; however, another thin connecting vessel between the afferent and efferent vessels became apparent. To avoid the recanalization of the aneurysm from the efferent side, the afferent vessel was embolized more proximally with 7 detachable coils, up to 5 mm in diameter and 15 cm in length (Target; Stryker). RCA angiography showed the disappearance of the aneurysm and CAF (Fig. 1C). The aneurysm was also not demonstrated on PA angiography.

After the procedure, the patient's symptoms disappeared. Two days later, echocardiography showed minimal pericardial effusion; thus, the pericardial drainage tube was removed. Echocardiography and contrast-enhanced CT confirmed the disappearance of the enhancement in the aneurysm (Fig. 1D). The patient remained symptom free at the 8-month follow-up.

Discussion

In angiographic studies, CAF is reported to have a prevalence of 0.2% [1]. The afferent vessels arise from the RCA in 50%, and left coronary artery in 42% of patients. The efferent vessels drain into the right atrium in 41%, PA in 17%, left ventricle in 3%, and superior vena cava in 1% of patients [5]. Most cases of CAF are single communications, but some cases of multiple fistula drainage sites have been reported [6]. Congestive heart failure, myocardial ischemia, infectious endocarditis, pulmonary hypertension, atrial fibrillation, and aneurysmal rupture are reported as the complications of CAF. The clinical presentation of CAF is mainly dependent on the severity of the left-to-right shunt and the majority of patients are asymptomatic [7]. Said reported that aneurysmal formation was found in 14% of patients with CAF and cardiac tamponade due to aneurysmal rupture was found in 2% of these patients [8].

Surgery has been the traditional therapeutic modality for closure of CAFs. However, TAE is less invasive than surgery and is now widely performed, although the early effectiveness, morbidity, and mortality for both approaches have been reported to be similar [6]. Recently, elective TAE for CAFs with unruptured aneurysms has also been reported [9,10]. Iwasawa et al. reported a case of CAF with a ruptured aneurysm, which was treated by TAE in emergency [4]. In their case, surgery was eventually required 2 weeks after TAE because of recanalization of the embolized site. To our knowledge, there has been no report of CAF with ruptured aneurysm treated using TAE alone.

In our patient with cardiac tamponade due to the ruptured aneurysm, surgery might have been a choice; however, our multidisciplinary team decided to treat her using TAE, because she was at an advanced age, her vital signs were stable after pericardiocentesis, and the CAF was anatomically simple, with a single communication (as seen on the CT and angiography). Technically, the coaxial combination of the 6-Fr guiding catheter, 4.2-Fr catheter, and microcatheter was useful to engage the RCA and to avoid microcatheter kick back during the embolization, resulting in complete isolation of the aneurysm by afferent and efferent vessels embolization. The procedure was successful, with no complications or recanalization.

In conclusion, TAE can be an alternative to surgery for CAFs, even in cases with ruptured aneurysms, if the patients' condition is stable and the CAF is anatomically simple. TAE is less invasive than surgery; thus, it is particularly suitable for elderly patients, as shown in our case. Multidisciplinary discussion is mandatory to decide the indication of TAE in the treatment of this fatal disease.

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