

Cardiac tamponade due to ruptured coronary-pulmonary artery fistula aneurysm: a case report

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Background

Coronary artery fistulas are rare and most commonly asymptomatic; however, they can become enlarged and rupture in some cases.

Case summary

We report a case of a 51-year-old woman who was brought to our hospital unconscious in an ambulance. Cardiac tamponade caused by the rupture of an aneurismal coronary-pulmonary artery fistula (CPAF) was detected by contrast-enhanced computed tomography and confirmed by invasive coronary angiography. Due to prompt diagnosis and subsequent surgical intervention, the patient's condition was rapidly improved, and she was discharged from the hospital.

Discussion

Coronary-pulmonary artery fistula aneurysm rupture requires rapid diagnosis and treatment, and thus, in cases with cardiac tamponade and coronary aneurysm, CPAF aneurysm rupture should be considered.

Keywords

Case report • Cardiac tamponade • Coronary artery fistula • Aneurysm

Learning points

- Three-dimensional computer tomography is useful for morphological evaluation of coronary artery fistulas.
- Coronary angiography should be considered in order to clarify the situation in detail if the patient's vitals are stable.
- We should consider a ruptured coronary artery fistula if cardiac tamponade and coronary aneurysm are observed.

Introduction

Coronary artery fistula is defined as a precapillary connection between a coronary artery and either of the low-pressure heart chambers or the pulmonary artery (PA).¹ Although most commonly asymptomatic, it can become enlarged and rupture. In such cases, emergency surgery is required. Herein, we report a case of cardiac tamponade due to ruptured coronary-pulmonary artery fistula (CPAF) aneurysm.

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Timeline

9:00	Onset of unconsciousness, ambulance call.
9:40	Arrival to our hospital. Echocardiography showed significant pericardial effusions. We initiated fluid replacement and pressor drugs.
10:00	Contrast-enhanced computed tomography (CT) revealed large aneurysms in front of the pulmonary artery.
11:00	Blood pressure declined despite medications, and we performed intubation and pericardiocentesis.
12:00	Coronary angiography showed a coronary-pulmonary artery fistula aneurysm.
13:30	Surgical treatment involving resection of the coronary artery aneurysm.
Post-operative day (POD) 1	Extubation and rehabilitation.
POD 7	Contrast-enhanced coronary CT showed patency of the coronary artery and bypass graft.
POD 13	Discharge from our hospital without any symptoms.
1-year after surgery	Regular follow-up.

Case presentation

A 51-year-old woman had lost consciousness while working at a pachinko parlour. She was a never-smoker and had no relevant medical history. She was transported to our hospital by ambulance and had regained consciousness somewhat but was still in somnolent state with limited orientation. On admission, the systolic blood pressure was 92 mmHg, the heart rate was increased at 98 b.p.m., and the Glasgow coma scale score was E3V4M6; she was in a pre-shock state. Blood testing revealed normal findings, including normal troponin T levels. Transthoracic echocardiography revealed a significant pericardial effusion, but the wall motion and valve function were normal. Chest X-ray showed enlargement of the cardiac silhouette. We suspected aortic dissection because of the pericardial effusion and performed three-dimensional contrast-enhanced computed tomography (CT). It revealed two large aneurysms (27 and 31 mm in size) on the anterior wall of the PA (Figure 1). We suspected CPAF with aneurysm. The blood pressure continued dropping despite the use of a vasopressor. Therefore, pericardial puncture was performed after endotracheal intubation; 400 mL of blood was drained and the blood pressure increased. Subsequently, we performed coronary angiography to determine the definitive diagnosis and the possibility of endovascular treatment.

Coronary angiography showed an aneurysm of a CPAF originating from the left anterior descending artery (LAD) (Figure 2). The right coronary artery was normal. Although leakage of contrast from the aneurysm was not confirmed, we suspected that the cardiac tamponade was caused by rupture of the CPAF aneurysm. Regarding the

treatment options, we considered that the blood flow from the PA side could not be blocked with a covered stent and that coil embolization would be necessary if we selected endovascular treatment. Thus, we agreed that surgical aneurysm removal was desirable. The patient was transferred to an operating room 4 h after symptom onset.

Intraoperatively, two coronary aneurysms were observed, 25 and 30 mm in size. One of the aneurysms, arising from the LAD and extending to the PA, was slightly bleeding (Figure 3). We made a small incision in the LAD and inserted a 1.5 mm probe to identify the LAD. Next, we resected the aneurysms and closed the fistula, suturing as close as possible to the LAD and PA. Subsequently, a longitudinal incision was made at the anterior PA wall and the outflow site of the CPAF was closed with suture. Considering the possibility of decreased blood flow in the LAD after the suture, left internal thoracic artery-to-LAD bypass was performed. The histological examination of the resected specimen showed arteriosclerotic changes in the form of intimal fibrous thickening. The elastic fibres were ruptured and lost, the stroma was oedematous, and mixed inflammatory cell infiltration was observed. It was inferred that the aneurysm formed due to arteriosclerotic changes in the vessel wall.

The patient was extubated 1 day after the operation. Contrast-enhanced coronary CT angiography performed on post-operative Day 7 showed a patent bypass and no narrowing of the LAD. The patient was discharged on hospital Day 14 and has had no symptoms 1 year after the operation.

Discussion

We reported a case of cardiac tamponade due to ruptured CPAF aneurysm. Coronary artery fistula was first described in 1865.² Although its frequency is 0.3–0.8% during coronary angiography,³ it reaches 4–15% during autopsy in cases of sudden death of young adults, suggesting that it is associated with sudden death.⁴ The origin of coronary artery fistulas is considered to be congenital, with no racial or gender differences.⁵ In 19–26% of cases of large left-to-right shunts due to the fistula, pulmonary hypertension, heart failure, rupture, and thrombus formation are seen,⁶ but the probability of aneurysm rupture is not clear.

According to a published case series by Ishii *et al.*,⁷ among 23 cases of coronary artery aneurysm rupture in Japan, 96% (22/23) of the patients had an aneurysm diameter of 3 cm or larger, and 96% (22/23) were women. The most common symptoms were loss of consciousness (39%, 9/23) and upper back pain (39%, 9/23). In the present case, the patient was also a woman and the presenting symptom was loss of consciousness. We could not confirm the presence of other symptoms as the patient was unconscious on arrival.

It was reported that coronary artery aneurysm rupture occurs mainly in postmenopausal middle-aged women, and the mechanism by which coronary fistulas form aneurysms is presumed to be related to the turbulence and arteriosclerosis caused by inflammation, meandering, and narrowing of the blood vessels.⁸

Three-dimensional CT is useful for morphological evaluation of coronary artery fistulas.⁹ In our case, we could follow the shunt blood vessel from the LAD to the PA by carefully following the coronary artery fistula. If an aneurysm is found around the coronary artery, it is

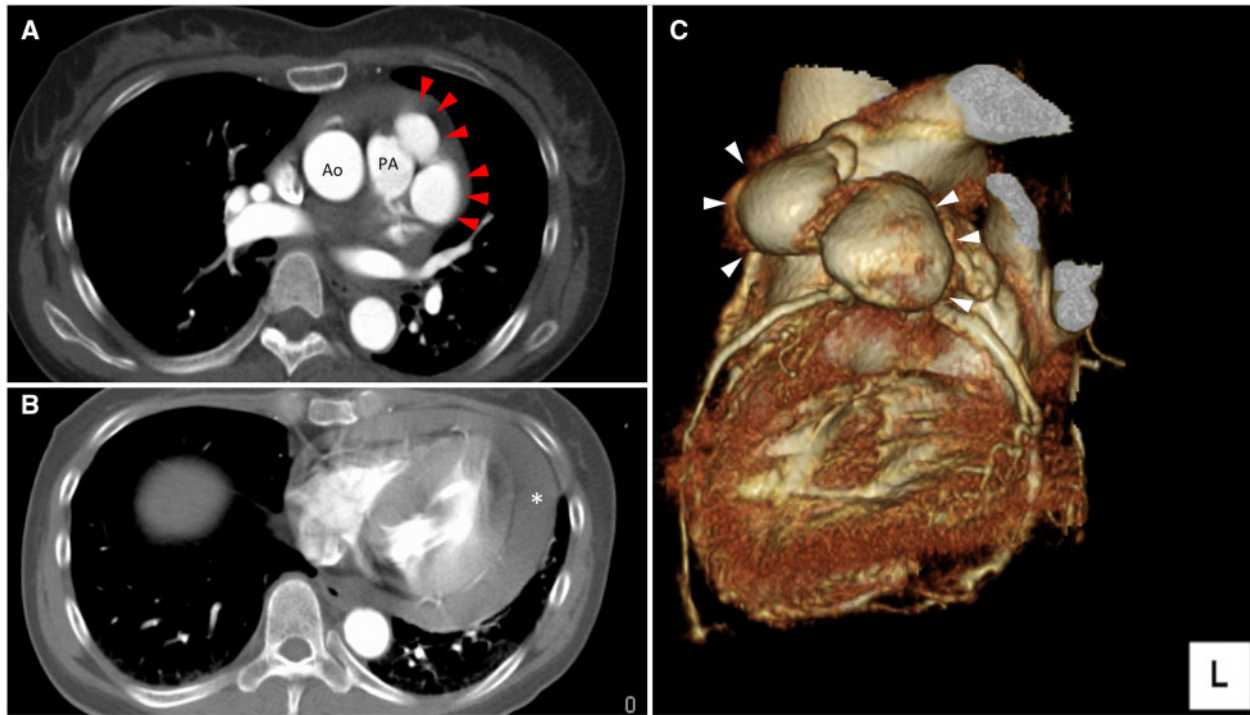


Figure 1 (A and B) Contrast-enhanced computer tomography. (C) Three-dimensional computer tomography. It revealed two large aneurysms (of 27 mm and 31 mm, respectively) on the front of the pulmonary artery. Pericardial effusion was detected. Ao, aorta; PA, pulmonary artery.

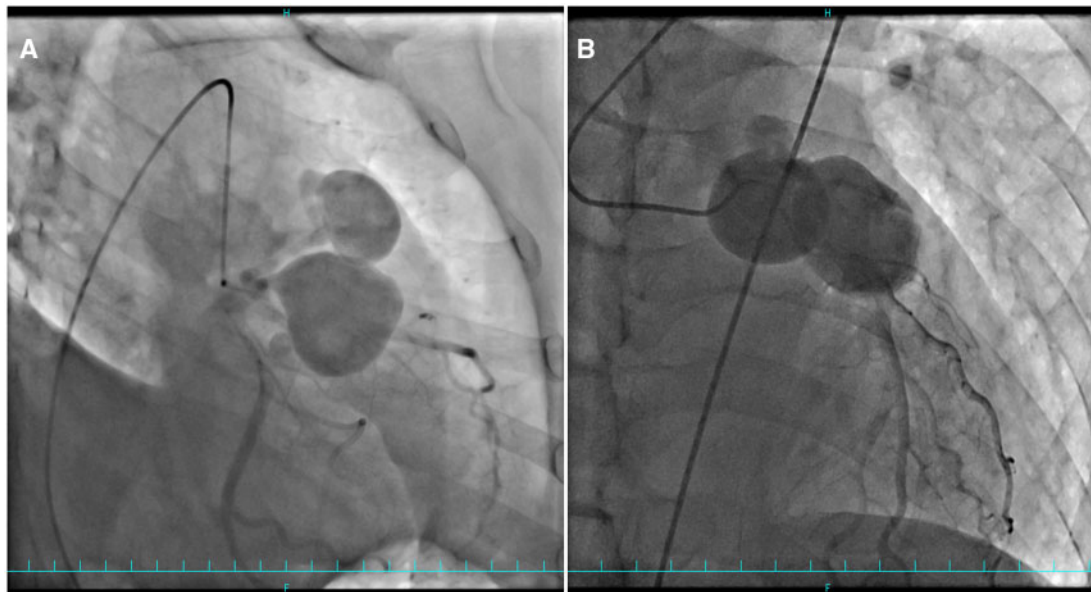


Figure 2 (A and B) Coronary angiography showed aneurysm formation of the coronary artery fistula from the left anterior descending artery to the pulmonary artery.

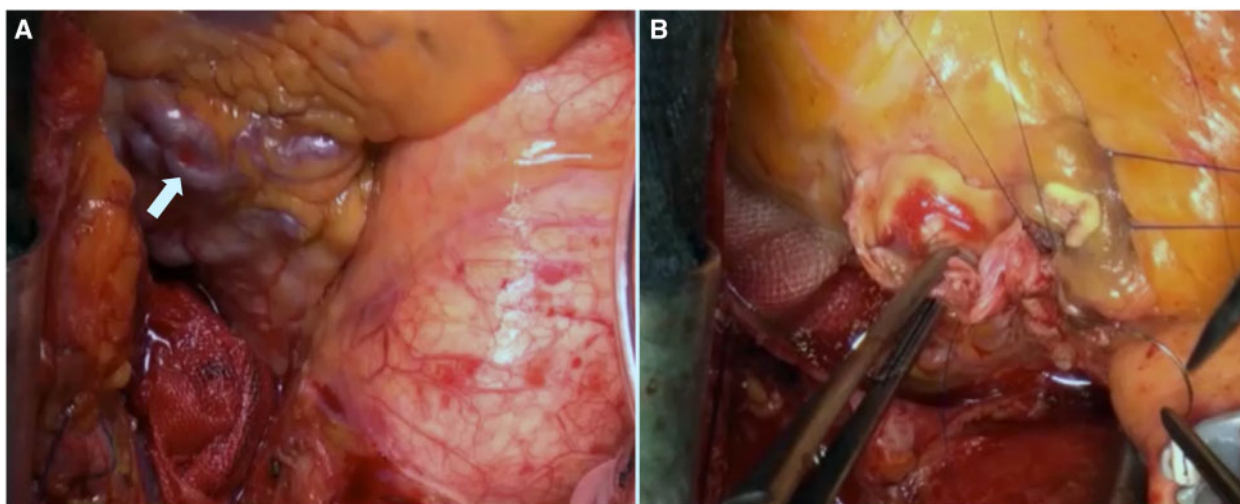


Figure 3 (A and B) During surgery, 25 mm and 30 mm coronary aneurysms were observed and one of them was slightly bleeding. It arised from left anterior descending artery and continues to pulmonary artery.

suspected to be formed due to the coronary artery fistula, and if the vitals are stable, it is considered desirable to perform coronary angiography before surgery because it can provide detailed information. For example, whether open-heart surgery or coil embolization is more desirable.

Regarding the surgical indications for coronary artery fistulas, Soji and Masahiro¹⁰ reported in 1973 that surgical treatment was indicated in case of: (i) asymptomatic fistula with left–right shunt rate of 30% or more; (ii) ischaemic changes on electrocardiography; (iii) advanced pulmonary hypertension or heart failure; (iv) history of bacterial endocarditis; and (v) risk of rupture due to enlarged aneurysms. As there have been many recent reports of ruptured aneurysms 3 cm or larger in size, it is considered that surgery may be indicated for aneurysms ≥ 3 cm to prevent rupture.^{8,11}

The treatment of coronary artery fistulas includes surgical excision and catheterization. Surgical treatment is advantageous in cases of high fistula flow and meandering, and when distal coronary artery bypass is required. Coil embolization is used for endovascular treatment.⁴ Notably, the efficacy and mortality rates in both treatments are similar.¹² There is also a report of covered stent treatment for coronary artery fistulas.¹³ However, this treatment seems difficult in case of bleeding from the aneurysm because the blood flow needs to be controlled from both sides of the fistula. There are also cases of aneurysms that have re-ruptured within a few days after surgical treatment or while waiting surgery. Therefore, prompt surgical treatment is desirable when aneurysm rupture is suspected.¹²

In conclusion, we reported a case of cardiac tamponade caused by a ruptured CPAF aneurysm. This condition requires rapid diagnosis and treatment. Hence, one should consider this condition if cardiac tamponade and coronary aneurysm are observed.

Lead author biography



Sadahiro Hijikata was born in Osaka in 1988. He became an interventional cardiologist because he felt working alongside the heart team to help patients was greatly rewarding. He graduated from Oita University in 2012. He became junior resident in Showa General Hospital from 2012 to 2014, and then a senior resident in cardiology, Tokyo Medical and Dental University Hospital from 2014 to 2015. After working at Japanese Red Cross Musashino

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Supplementary material

Supplementary material is available at *European Heart Journal - Case Reports* online.

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Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as [Supplementary data](#).

Consent: The author/s confirm that written consent for submission and publication of this case report including image(s) and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: none declared.

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