

Cardiac Tamponade Due to Rupture of Coronary Artery Fistulas With a Giant Aneurysm Containing a Free Floating Ball Thrombus: A Case Report

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Abstract

A 76-year-old woman was admitted to our hospital with cardiac tamponade. Chest computed tomography revealed a large tumor adjacent to the pulmonary artery, the center of which was demonstrated as a high density area without contrast medium. Coronary angiography revealed a large aneurysm consisting of coronary artery fistulas. An afferent vessel connected tangentially to the surface of the aneurysm, so blood flowed around along the inner wall of the aneurysm. A filling defect was demonstrated in the center of the aneurysm that indicated a thrombus. The cause of cardiac tamponade was seemed to be bleeding of the coronary artery fistulas. Coil embolization was performed, but recanalization occurred. Therefore, ligation of the fistulas and aneurysmal resection were performed using a cardiopulmonary bypass. A fresh floating thrombus was found in the aneurysm.

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Key Words

■ Aneurysms ■ Cardiac tamponade
■ Coronary vessels (coronary artery fistula)

■ Thrombosis (floating thrombus)

INTRODUCTION

Coronary artery fistula is a rare anomaly that consists of a communication between the coronary arteries and a cardiac chamber or great vessel, and is usually asymptomatic. The clinical presentations, which are mainly dependent on the severity of the left-to-right shunt, include fatigue, dyspnea, and angina. Potential complications include pulmonary hypertension, congestive heart failure, myocardial ischemia due to coronary steal phenomenon, infec-

tive endocarditis, and rupture or thrombosis of the fistulas.¹⁾

We report a case of rupture of coronary artery fistulas with a giant aneurysm containing a free floating ball thrombus manifesting as cardiac tamponade.

CASE REPORT

A 76-year-old woman with a history of hypertension was admitted to our hospital with dizziness and hypotension. On admission, her blood pressure

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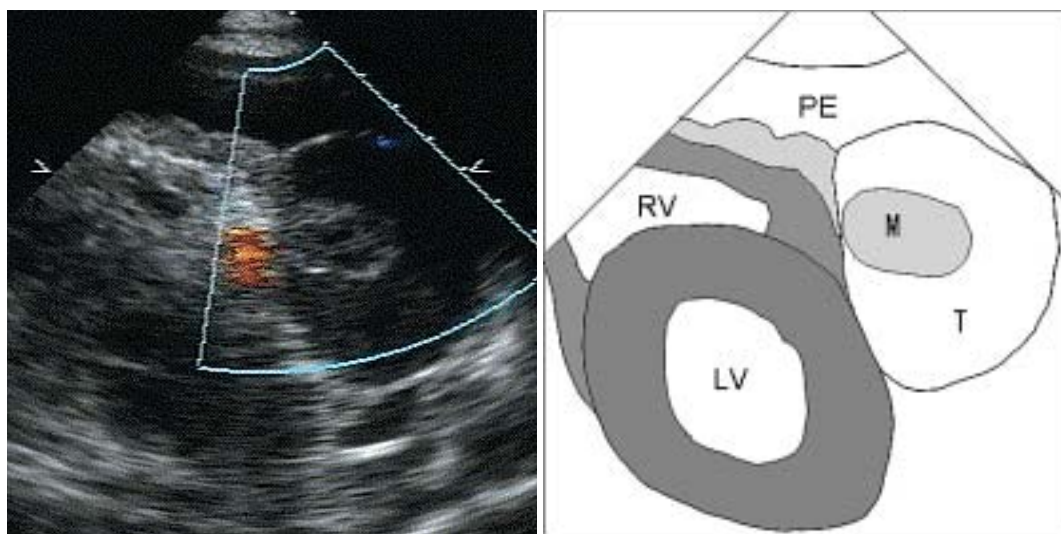


Fig. 1 Doppler echocardiogram (left) and schematic representation (right) from the parasternal short-axis view

Echo free space and a large tumor in front of the basal left ventricle were seen. A 2 cm mass was detected in the tumor.

LV = left ventricle; RV = right ventricle; PE = pericardial effusion; T = tumor; M = mass.

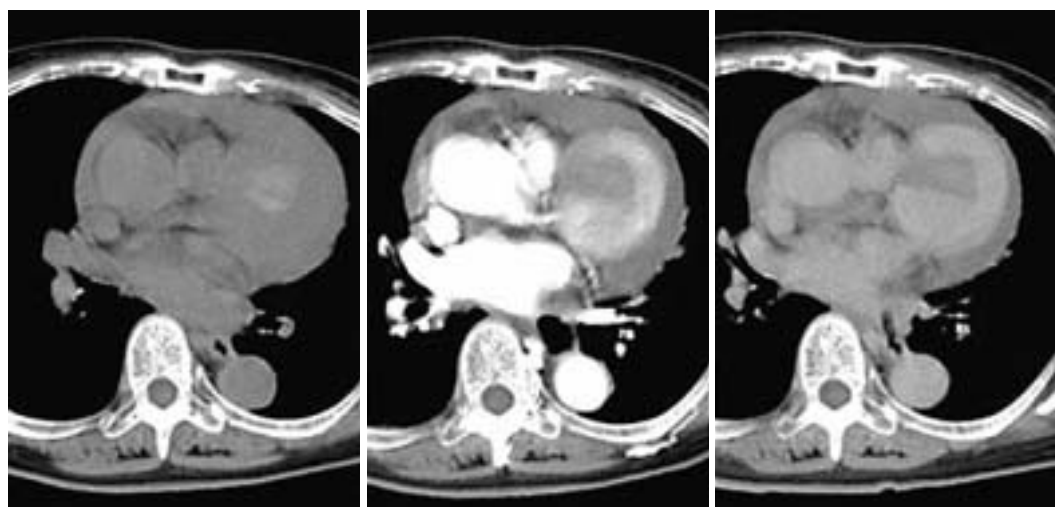


Fig. 2 Chest computed tomography scans

Without contrast medium (left), early phase (middle) and late phase (right) with contrast medium, showing the large tumor adjacent to the pulmonary artery.

was 92/68 mmHg and pulse rate was 106 beats/min. Her physical examination was remarkable for a continuous murmur of Levine II at the left upper sternal border and jugular venous distention. Paradoxical pulse was not apparent. Echocardiography revealed massive pericardial effusion and collapse of the right ventricle during early diastole. A large (5 × 5 cm in diameter) tumor was

detected in the front of the basal left ventricle. Abnormal blood flow signals and a 2 cm mass were detected in the tumor (Fig. 1). Chest computed tomography (Fig. 2) revealed the tumor adjacent to the pulmonary artery. The center of the tumor was demonstrated as a high density area without contrast medium. The tumor was enhanced by contrast medium like the great vessels except for the center

of the tumor. The computed tomography number of the pericardial effusion was 48.6, indicating hemo-pericardium. Chest T1-weighted magnetic resonance imaging (**Fig. 3**) showed the left anterior descending coronary arteries below the tumor, but could not detect the continuity between the tumor and the coronary arteries.

We considered that the cause of the cardiac tamponade was bleeding of the tumor. Urgent coronary angiography established the differential diagnosis from malignancy. Left coronary angiography (**Fig. 4**) showed a large aneurysm arising from the proximal part of the left anterior descending artery and draining into the pulmonary artery. An afferent vessel ran straight and connected with the bottom of the aneurysm tangentially, so contrast medium flowed along the inner wall of the aneurysm. A round filling defect was demonstrated in the center of the aneurysm. Smaller fistulas, which had not formed aneurysms, were also detected between the right coronary artery and the pulmonary artery. A

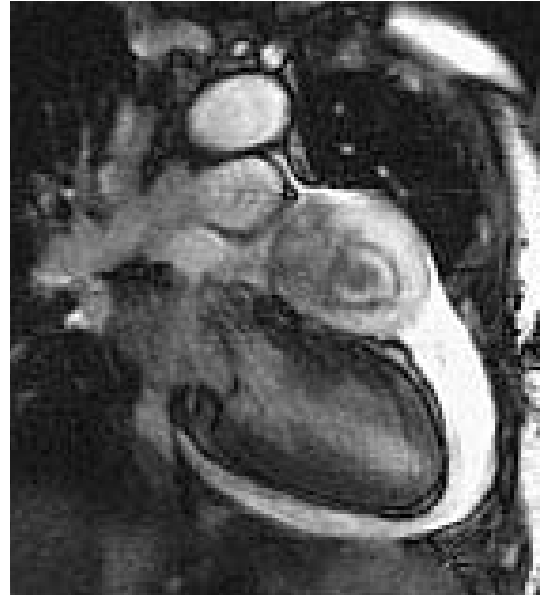


Fig. 3 Chest T1-weighted magnetic resonance image

The tumor is located in front of the basal left ventricle with pericardial effusion.

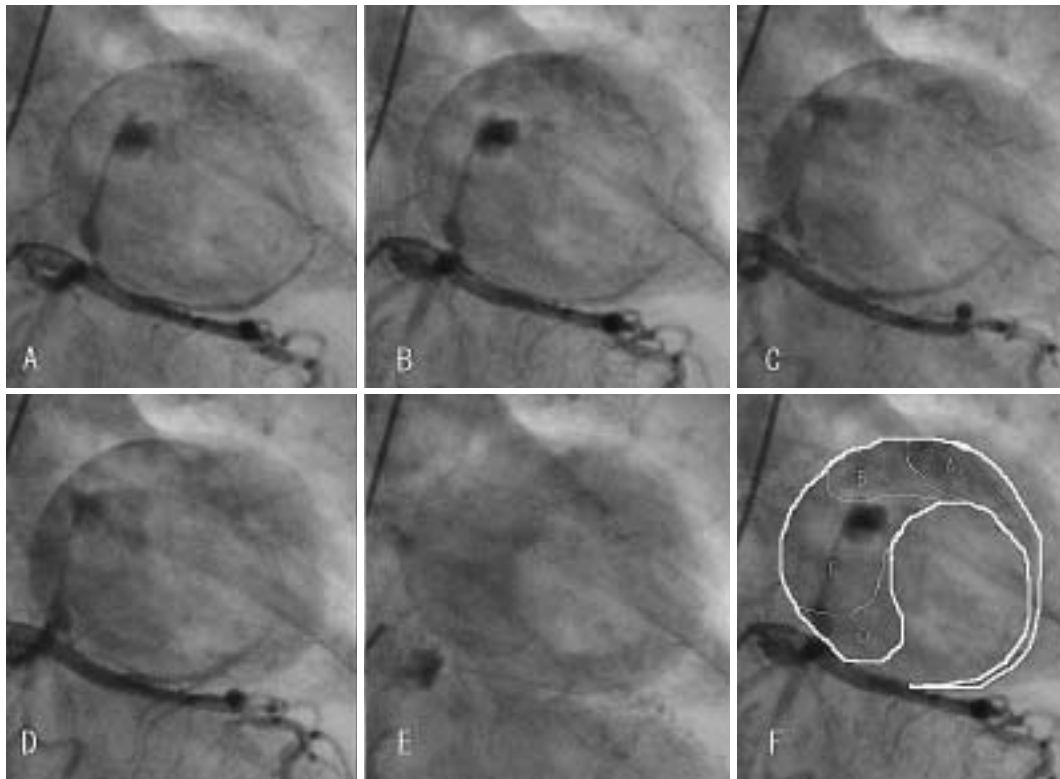


Fig. 4 Left coronary angiograms

A large aneurysm arose from the proximal part of the left anterior descending artery. An afferent vessel was straight and connected with the bottom of the aneurysm tangentially, and contrast medium showed counterclockwise flow along the inner wall of the aneurysm (A, B, C, D in order). A round filling defect was seen in the center of the aneurysm (E), schema of blood flow in the aneurysm (F).

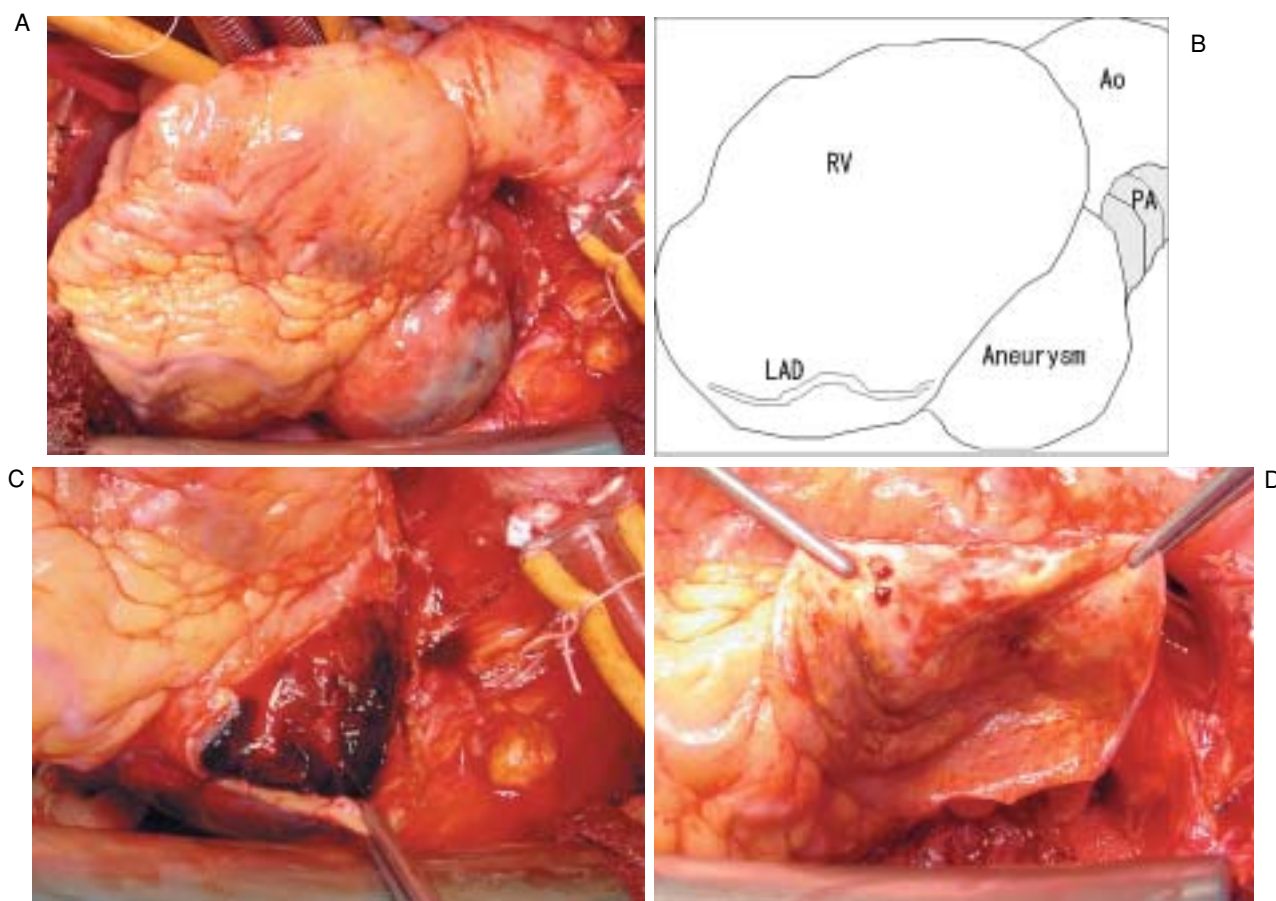


Fig. 5 Operative photographs (A, C, D) and schematic representation (B)

A: A large (5 cm in diameter) aneurysm of coronary artery fistula was located between the pulmonary artery and left anterior descending coronary artery.

B: RV = right ventricle; Ao = ascending aorta; PA = pulmonary artery; LAD = left anterior descending coronary artery.

C: Soft thrombus was detected in the aneurysm.

D: Aneurysm inner wall.

significant rise in oxygen saturation was observed in the pulmonary artery, and the left-to-right shunt ratio was 19%. The clinical diagnosis was cardiac tamponade due to rupture of the coronary artery fistulas with a giant aneurysm containing a floating ball thrombus.

We performed coil embolization of the fistulas and drainage of 300 ml bloody pericardial effusion, when her blood pressure was normalized (134/60 mmHg). Two weeks after the treatment, coronary angiography revealed recanalization of the embolization site. We performed ligation of the fistulas originating from the right and left coronary arteries, and resection of the aneurysm using cardiopulmonary bypass (**Fig. 5**). The resected aneurysm showed smooth arterial wall and no atherosclerotic change such as calcification.

Histological study of the aneurysm wall revealed trilaminar structure and inflammatory cellular infiltration in the adventitia. The rupture site could not be detected. A fresh free floating ball thrombus (**Fig. 6**) was found in the aneurysm. The postoperative course was uneventful, and she was discharged home.

DISCUSSION

Coronary artery fistulas are identified incidentally in 0.3–0.8% of routine cardiac angiographic series.¹⁾ The right coronary artery is the site of the fistula in about 55% of cases, the left coronary artery in about 35%, and both coronary arteries in 5%.¹⁾ Aneurysmal formation is present in 19–26%,^{2,3)} and rupture of the fistulas, pericardial effusion, and sudden death are the presenting fea-

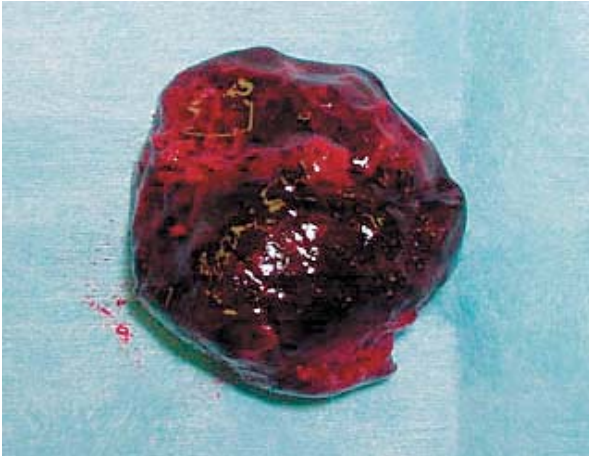


Fig. 6 Photograph of the free-floating ball thrombus in the aneurysm

tures.^{4,5)} Thrombosis of the fistulas is also recognized. However, this case involved an unusual free floating ball thrombus in the aneurysm of the coronary artery fistulas. In this case, an afferent vessel connected tangentially to the surface of aneurysm, so blood flowed around along the inner wall of the aneurysm. The free floating ball thrombus was formed in the center of the aneurysm where the blood flow stagnated. We were misled by the presence of this free floating ball thrombus. The thrombus was very fresh, so appeared as high density on computed tomography without contrast medium and the aneurysm was well enhanced, mimicking a malignant tumor in which bleeding occurred.

Coronary angiography was most valuable for the diagnosis in this case. Cardiac catheterization may be indispensable for the diagnosis and hemodynamic evaluation of coronary artery fistulas. Various cardiac imaging modalities are utilized for adjunct diagnostic evaluation. Two-dimensional echocar-

diography can usually detect an enlarged feeding vessel with an abnormal flow pattern.^{6,7)} Multiplanar transesophageal echocardiography also accurately defines the origin, course, and drainage site of the fistulas.⁸⁾ Ultrafast computed tomography or magnetic resonance imaging with contrast medium can evaluate the course of abnormal tortuous blood vessels using multiplanar reconstruction.^{9,10)} However, several limitations¹⁾ have been described with these imaging techniques. Various modalities should be used to establish the diagnosis.

Most adult patients with coronary artery fistulas remain free from symptoms for long periods.¹¹⁾ Small fistulas may close spontaneously, especially in pediatric patients.¹²⁾ The management for asymptomatic patients is still controversial. Patients with complications need to undergo surgical or percutaneous intervention. Surgical closure¹³⁾ and percutaneous transcatheter closure¹⁴⁾ are both safe and effective. Transcatheter closure has been proposed as the initial choice, even if staged procedure such as surgical intervention is ultimately required.¹⁵⁾ In our patient, recanalization was recognized after coil embolization, possibly because the afferent vessel of the aneurysm was straight and tapering, and blood flow velocity in the afferent vessel was relatively high. If recanalization occurs after transcatheter closure, we should not hesitate to perform the surgical management of a patient with severe complications like this case.

The present case of cardiac tamponade due to rupture of coronary artery fistula with a giant aneurysm containing a giant thrombus floated shows that early diagnosis and early treatment is important to manage this very rare, but severe complication.

要 約

巨大瘤を伴う冠動脈瘻の破裂により心タンポナーデをきたした1例：
瘤内球状浮遊血栓の合併

岩澤 祐二 北村 豊 日隈菊比呂
小野 文明 井元 清隆 木村 一雄

症例は心タンポナーデを呈した76歳、女性。胸部コンピューター断層撮影(CT)で血性心嚢液貯留と肺動脈に接する径5cmの腫瘍が認められた。腫瘍中心部は単純像で高濃度を呈し、腫瘍内部

の辺縁寄りには濃く造影された。冠動脈造影検査の結果、この腫瘍は冠動脈瘻の巨大瘤と診断され、心タンポナーデの原因は瘻破裂による出血と考えられた。瘤への流入血管は瘤壁表面を直線的に走行し接線方向に開口しており、瘤内の血流が内壁に沿って円周状に渦巻くように観察された。血流の滞る中心部に島状の陰影欠損が認められ、単純CTで高濃度を呈したものは浮遊血栓と思われた。コイル塞栓術後に心嚢ドレナージが行われたが、塞栓部位の再疎通が認められ、人工心肺下に冠動脈瘻結紮術と瘤切除術が施行された。瘤内から比較的新鮮な球状浮遊血栓が摘出された。冠動脈瘻の破裂はまれであり、さらに本例は冠動脈瘻の巨大瘤と瘤内の特異な血流パターンのために形成された球状浮遊血栓を観察しえた貴重な症例であった。

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