Clinical Case Report

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Bilateral coronary artery fistulas with a giant coronary aneurysm complicated by aneurysm rupture

A case report

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Abstract

Rationale: Coronary artery aneurysms and fistulas are not rare conditions in clinical practice, but bilateral fistulas with a giant coronary aneurysm in just one person are quite rare.

Patient concerns: We report a case of a 66-year-old woman with these 2 coronary abnormalities accompanied with a huge mediastinum mass.

Interventions: The giant aneurysm was ligated and the mass was resected which was proved to be an organized hematoma finally.

Outcomes: The patient was discharged soon with no complications.

Lessons: The best treatment of giant coronary aneurysm is not clear because of its rarity, surgical resection may be the right procedure for the potential serious complications like this case.

Abbreviations: CT = computed tomography, LAD = left anterior descending artery.

Keywords: aneurysm rupture, coronary artery aneurysm, coronary artery fistula

1. Introduction

Coronary artery fistula is a congenital or acquired abnormality of coronary anatomy which consists of connections between a coronary artery and cardiac chambers, a coronary vessel, or an intrathoracic vessel.[1] And coronary artery aneurysm is a coronary artery dilatation that exceeds the diameter of normal adjacent segments or the diameter of the patient's largest coronary vessel by 1.5 times. It is called giant coronary artery aneurysm, if the diameter is more than 20 mm.[2] Coronary artery aneurysms and fistulas are not rare conditions in clinical practice by angiography, usually do not need any special treatment, but in some cases they can cause serious complications.

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2. Case report

A 66-year-old woman was admitted to our hospital for mediastinum tumor that had been detected upon computed tomography. She had no heart symptoms, but a continuous murmur was heard over the precordium near the left sternal border. The electrocardiogram was normal. Transthoracic echocardiography was performed demonstrating normal left ventricular size and function, normal wall motion, and no pericardial effusion. Chest computed tomography (CT) with contrast indicated a huge round mass in the cavum pericardii adjacent to the left ventricular, with the dimension of 68 × 74 mm (Fig. 1 A and B).

Before surgery we did coronary angiography that revealed bilateral coronary artery fistulas and a giant coronary aneurysm. The left main and the left anterior descending artery (LAD) were dilated, and the giant aneurysm (also fistula) was arising in proximal LAD ending in the pulmonary artery (Fig. 2A–C), we could also see a branch coming from the midportion of LAD to the round mass with contrast enhancement (red arrow). The second fistula was arising in the ostium of right coronary artery and terminating in the pulmonary artery (Fig. 2E and F). The huge mass was resected and the proximal and distal openings of the giant aneurysm were ligated. The histological analysis of the resected tissue showed a lot of erythrocytes, fibrosis, hyalinization, calcification, regional lymphocyte, and plasmocyte infiltration (Fig. 3). The patient was discharged soon with no complications.

Ethical approval was not required for this case report as it did reveal the patient's name and privacy. Informed consent for the publication of this case report was obtained from the patient herself.

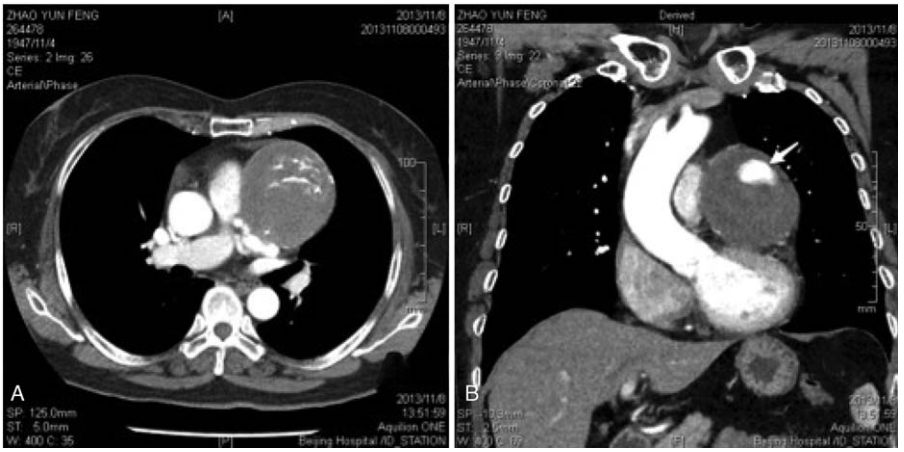

Figure 1. Chest computed tomography scanning demonstrated a huge round mass with contrast enhancement (arrow). (A) the transverse section of the mass; (B) the coronal section of the mass.

3. Discussion

Coronary artery aneurysms and fistulas are not uncommon in clinical practice by angiography, coronary artery aneurysms are detected in 0.15% to 4.9% of patients who undergo coronary angiography,[3] and coronary artery fistulas are detected in about 0.1% patients.[4] But bilateral fistulas with a giant coronary aneurysm in just 1 person are quite rare; moreover, this patient progressed with a round mass in the cavum pericardii.

Most fistulas are incidentally identified, as they often do not cause hemodynamically relevant shunt or symptoms, but in some cases with huge shunt can cause chest pain or heart failure on the basis of coronary steal phenomenon.[5]

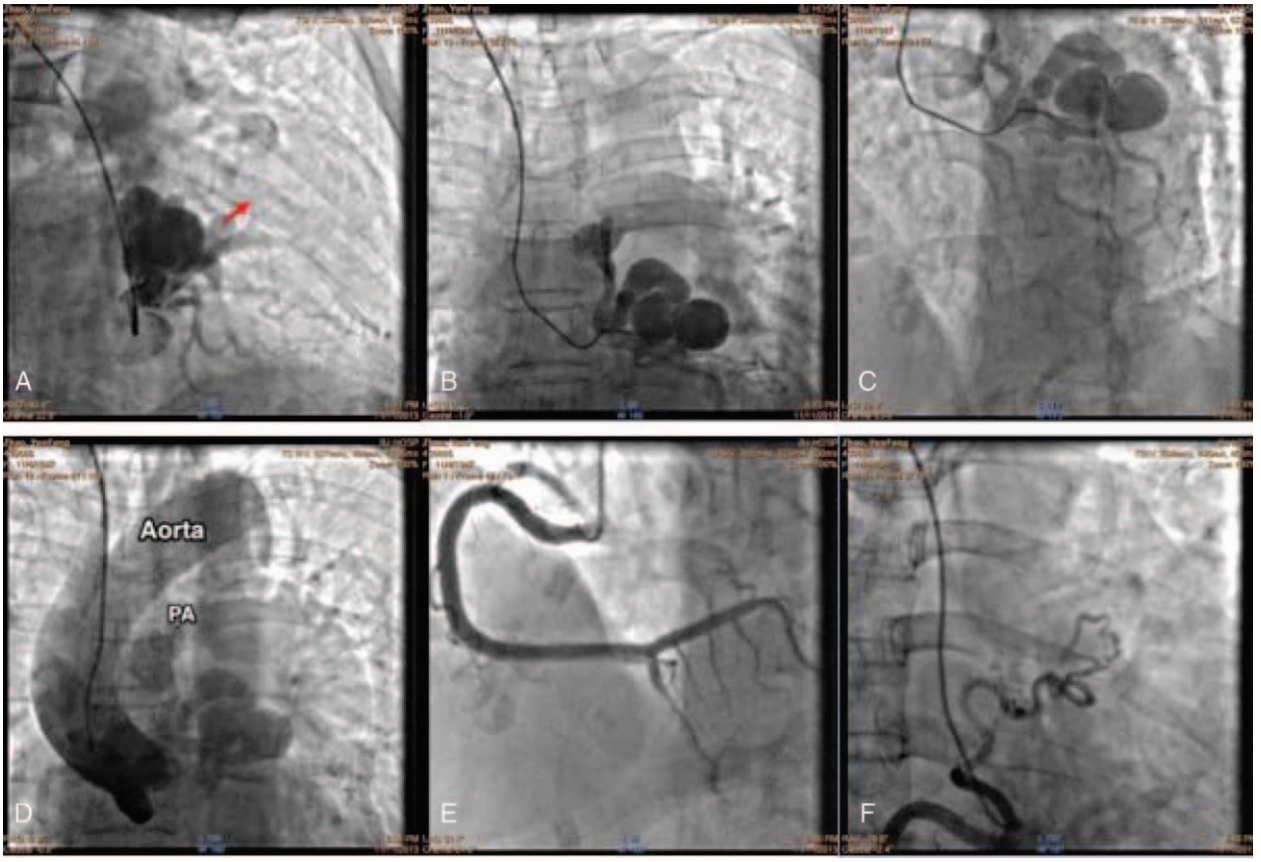

Figure 2. Coronary angiography showed a giant fistula arising in left anterior descending artery ending in the pulmonary artery (A–C), the red arrow showed a branch into the mass from LAD (A); ascending aorta angiography showed the relationship of aorta, pulmonary artery, and the giant aneurysm (D); the second fistula arising in right coronary artery and terminating in the pulmonary artery (E and F). LAD = left anterior descending artery.

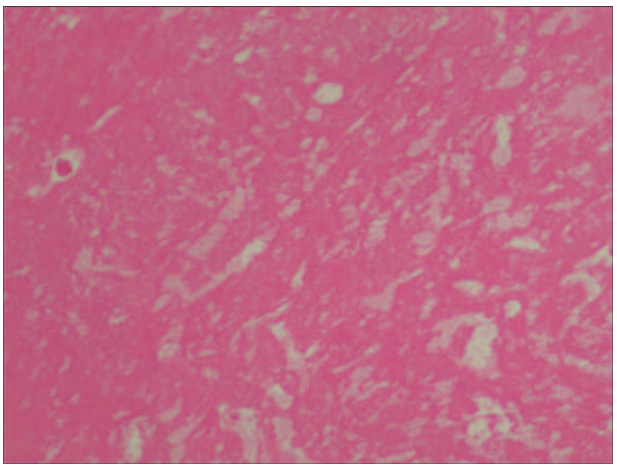

Figure 3. Histological analysis of the resected tumor. Hematoxylin and eosin staining.

The common reasons for 5 coronary aneurysm are atherosclerosis, Kawasaki disease, congenital, trauma, angioplasty, arteritis (including syphilis), mycotic, connective tissue disorders, and dissection.[3] This patient did not have any evidence of collagen vascular diseases, chest trauma, or other special diseases, coronary angiography showed no atherosclerosis lesions and all the vessels were very smooth, so the giant coronary aneurysm may have occurred as a result of high flow of blood from a high-pressure chamber (aorta) to a low-pressure chamber (pulmonary artery) due to the congenital fistula, just as Morita et al[6] reported previously. And coronary artery fistula is a possible reason for coronary aneurysm, but it is quite difficult to explain just one fistula causing giant coronary aneurysm and the other one not.

Patients with coronary artery aneurysms are usually asymptomatic, the potential serious complications include rupture, thrombosis, embolization, dissection, mechanical obstruction, and erosion into surrounding structures, especially for the giant aneurysms.[7,8] From the patient's medical history, CT scanning, and resected tissue histopathology, we thought the huge mass may have occurred due to the giant aneurysm rupture, enwrapped, and organization. Rupture of a coronary artery aneurysm is extremely rare, although it was reported in the literature that acute coronary aneurysm rupture caused cardiac tamponade, unconsciousness, and even death,[9,10] but chronic aneurysm rupture like this case has not been described previously. Until now the best treatment of giant coronary aneurysm is not clear because of its rarity, surgical resection may be the right choice for the potential serious complications like this case.

References

[1] Loukas M, Germain AS, Gabriel A, et al. Coronary artery fistula: a review. Cardiovasc Pathol 2015;24:141–8.
[2] Jha NK, Ouda HZ, Khan JA, et al. Giant right coronary artery aneurysm. J Cardiothorac Surg 2009;4:18.
[3] Syed M, Lesch M. Coronary artery aneurysm: a review. Prog Cardiovasc Dis 1997;40:77–84.
[4] Vavuranakis M, Bush CA, Boudoulas H. Coronary artery fistulas in adults: incidence, angiographic characteristics, natural history. Cathet Cardiovasc Diagn 1995;35:116–20.
[5] Andò G1, Ascenti G, Saporito F, et al. Multislice computed tomography demonstration of a coronary-to-pulmonary artery fistula. J Cardiovasc Med (Hagerstown) 2011;12:212–4.
[6] Morita H, Ozawa H, Yamazaki S, et al. A case of giant coronary artery aneurysm with fistulous connection to the pulmonary artery: a case report and review of the literature. Intern Med 2012;51:1361–6.
[7] Crawley PD, Mahlow WJ, Huntsinger DR, et al. Giant coronary artery aneurysms: review and update. Tex Heart Inst J 2014;41:603–8.
[8] Kumar G, Karon BL, Edwards WD, et al. Giant coronary artery aneurysm causing superior vena cava syndrome and congestive heart failure. Am J Cardiol 2006;98:986–8.
[9] Daneshvar DA, Czak S, Patil A, et al. Spontaneous rupture of a left main coronary artery aneurysm. Circ Cardiovasc Interv 2012;5:63–5.
[10] Kazuno K, Akasaka N, Kiyokawa K, et al. Ruptured aneurysm of coronary artery-to-pulmonary artery fistula. Asian Cardiovasc Thorac Ann 2012;20:324–6.