Case Report

Spontaneous rupture of a giant coronary artery aneurysm causing cardiac tamponade: A case report

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Summary A 62-year-old woman with a history of dyslipidemia and hypothyroidism was referred to our institution with syncope. Cardiac tamponade due to spontaneous rupture of a 50-mm aneurysm of the coronary artery was diagnosed by transthoracic echocardiography, enhanced computed tomography, and coronary angiography. Emergency surgery was performed, and despite developing postoperative complications such as acute renal insufficiency, the patient was discharged from hospital without sequelae 89 days later. Histological findings revealed cystic media degeneration, but neither significant atherosclerotic changes nor inflammatory cell infiltration. Although coronary artery aneurysms are comparatively rare and generally asymptomatic, those over 30 mm in diameter are considered to be at increased risk of rupture. A coronary artery aneurysm of about 50 mm ruptured in our patient, supporting this view.

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Introduction

Coronary artery aneurysms are quite rare, occurring in 0.15–4.9% of patients undergoing coronary angiography. Giant (>20 mm in diameter) coronary artery aneurysms are even more infrequent. Although most coronary artery aneurysms are asymptomatic, they can cause serious complications such as thromboembolism, rupture, and compression of adjacent organs. Most reported ruptured coronary artery aneurysms have been >30 mm in diameter. We describe a giant (50 mm in diameter) right coronary artery aneurysm that ruptured and presented as cardiac tamponade. A histopathological assessment showed cystic media degeneration, which probably also played a key role in the origin of the coronary artery aneurysm.

Case report

A 62-year-old woman with a history of dyslipidemia and hypothyroidism was referred to our institution with syncope. She had no history of Kawasaki disease or the
Figure 1  Enhanced computed tomography shows large spherical mass adjacent to the right heart and pericardial effusion, communicating proximally with right coronary artery ostium and distally with distal right coronary artery (A and B). Coronary angiography shows extremely large saccular aneurysm of the right coronary artery and a normal left coronary artery (C and D).

Figure 2  Aneurysm of about 50 mm in diameter originates from ostium of right coronary artery. Rupture site is located on rear wall of the aneurysm. No significant mural thrombus is evident in the aneurysm.

Figure 3  Histopathological findings of the excised aneurysm. Cystic media degeneration (A) and coronary dissection (B) are evident. HE, hematoxylin and eosin; EvG, elastic-Van Gieson.
skeletal abnormalities associated with Marfan syndrome. Although the femoral artery was palpable, blood pressure was impossible to measure and her level of consciousness was GCSE3V5M6. Auscultation detected diminished heart sounds but no significant murmur. Laboratory analyses showed elevated liver enzymes that were probably induced by shock. Chest radiography showed an abnormally prominent right heart border, and electrocardiography showed atrial fibrillation with a rapid ventricular response of around 200/min and no significant ST-T segment abnormalities. Transthoracic echocardiography revealed a large spherical mass adjacent to the right heart, echogenic pericardial effusion, and regional hypokinesia of the posteroinferior wall of the left ventricle. Enhanced computed tomography (CT) confirmed that a mass communicated proximally with the right coronary artery ostium and distally with the distal right coronary artery. Coronary angiography revealed an extremely large saccular aneurysm of the right coronary artery and a normal left coronary artery (Fig. 1). These findings indicated a diagnosis of cardiac tamponade due to rupture of a giant right coronary artery aneurysm, and emergency surgery was performed. Aneurysmectomy and coronary artery bypassgrafting proceeded through a median sternotomy under cardiopulmonary bypass established through cannulation of the left femoral vessels. After pericardiectomy, arterial bleeding was evident and a rupture site was located on the rear wall of the aneurysm, which was about 50 mm in diameter and originated from the ostium of the right coronary artery (Fig. 2). The proximal and distal portions of the aneurysm were ligated and the aneurysm was resected. The right coronary artery was reconstructed using a bypass saphenous vein graft to its distal portion. Although the patient was warmed, she remained in shock due to right heart failure despite catecholamine administration. The cardiopulmonary bypass was followed by the insertion of an intra-aortic balloon pump (IABP) and temporary pacing, but the patient developed acute renal failure due to prolonged shock. However, right ventricular wall motion gradually improved and she recovered from shock under catecholamine medication, IABP, and continuous renal replacement therapy. Although she developed hemorrhagic enteritis, she recovered conservatively and was discharged without sequelae on hospital day 89. A histopathological assessment of the excised aneurysm revealed cystic media degeneration and coronary dissection (Fig. 3). Coronary artery CT at 1 year after surgery showed a patent bypass graft and no new aneurysms. Transthoracic echocardiography showed good cardiac function.

Discussion

Coronary artery aneurysm is defined as a localized dilation exceeding the diameter of an adjacent normal segment by 50% [1]. Those with a diameter of >20 mm are referred to as giant coronary artery aneurysms [2]. The reported incidence of coronary artery aneurysm ranges from 0.15% to 4.9% of patients undergoing coronary angiography, and the frequency of giant coronary artery aneurysms is thought to be even lower. The causes of coronary artery aneurysms include atherosclerosis, Kawasaki disease, infection, trauma, and congenital conditions. However, the incidence varies geographically — atherosclerosis causes 50% of coronary artery aneurysms in Europe or North America, whereas Kawasaki disease is the main cause in Japan and China. Histopathology of resected specimens from our patient showed cystic media degeneration, but neither significant atherosclerosis nor inflammatory cell infiltration. Our patient did not have a family history or the specific skeletal abnormalities associated with Marfan syndrome. The numbers of reports of vascular lesions associated with cystic media degeneration in non-Marfan patients have recently increased and include aortic, carotid, and other visceral artery lesions [3—5]. Cystic media degeneration weakens the aortic media, which results in aneurysmal dilatation. Only one other report has described a similar patient without Marfan syndrome in whom cystic media degeneration of the coronary arterial wall caused a giant coronary artery aneurysm [6]. On the other hand, because the frequency of cystic media degeneration increases with age, and is associated with hypertension, it is considered as a normal feature of aging. However, our patient did not have a history of Kawasaki disease and a histopathological assessment of the aneurysm did not uncover any significant atherosclerosis or inflammatory cell infiltration. Thus, we considered that cystic media degeneration was associated with formation of the giant coronary artery aneurysm.

Most patients with coronary artery aneurysms are asymptomatic, but they can present with rupture, thromboembolism, and compressed surrounding structures. Although treatment for giant coronary aneurysms has not yet been standardized, patients with enlarging aneurysms should be treated regardless of whether or not they are symptomatic. In addition, the diameter of almost all ruptured aneurysms is >30 mm and thus such aneurysms should be surgically treated [7]. Our patient had been attending another hospital for hypothyroidism, and no prior images were presented that could have detected a coronary artery aneurysm. Thus, when the coronary artery aneurysm occurred and the rate at which it increased remain unknown. The coronary artery aneurysm could have been explored because it had rapidly grown. However, our patient had a 50 mm aneurysm at the time of rupture, so only the size of the aneurysm was sufficient to cause rupture. When such giant aneurysms (>30 mm in diameter) are discovered, we also suggest that they be treated to avoid rupture. Although we treated our patient by surgical resection, percutaneous approaches such as covered stents [8] and coil embolization [9] have also been applied. The size and shape of an aneurysm should be confirmed so that the appropriate therapeutic approach can be selected.

The 5-year survival rate of patients with aneurysms was 71% in one report that defined a coronary artery aneurysm as being over double the size of the normal coronary artery or more than 8 mm [10]. However, the prognosis of patients with giant coronary artery aneurysms (>20 mm in diameter) is unknown.

To diagnose an asymptomatic coronary artery aneurysm is extremely challenging and thus problematic. Case reports have described asymptomatic coronary artery aneurysms that presented as a cardiac murmur or chest X-ray abnormalities. These reports suggest the importance of a detailed physical examination and careful interpretation of chest X-ray findings.
References