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A successful surgical management for a thrombosed giant left coronary aneurysm with right ventricular fistula in a young patient

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Brief Report

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Abstract

Giant coronary aneurysm with ventricular fistula is rare. Due to the limited data from randomised clinical trials, therapeutic strategies for coronary aneurysms predominantly rely on on case series and anecdotal evidences. Reporting cases that provide practical experience in managing these aneurysms is therefore crucial. In this article, we report a rare case of a successful surgical management for a thrombosed giant left coronary aneurysm with right ventricular fistula, which is larger than any previously reported cases.

Coronary artery fistula refers to abnormal communications between coronary arteries and either the cardiac chambers or thoracic great vessels. Most coronary artery fistulas are congenital.[1] Coronary artery fistulas are extremely rare, constituting 0.2% to 0.4% of all congenital cardiac anomalies.[2] The most frequent complication of coronary artery fistula is the development of a coronary aneurysm, defined as localised coronary dilation, with a diameter exceeding 1.5 times that of a normal adjacent coronary artery. When its diameter exceeds 20 mm, it is classified as a giant coronary aneurysm. The overall prevalence of giant coronary aneurysm in the population is 0.02–0.2%.[3] A giant coronary aneurysm with ventricular fistula is even rarer. Due to the limited data from randomised clinical trials, therapeutic strategies for coronary aneurysms predominantly rely on case series and anecdotal evidences. Reporting cases that provide practical experience in managing these aneurysms is therefore crucial. In this article, we report a rare case of a successful surgical management of thrombosed giant coronary aneurysm with right ventricular fistula in a young patient, which is larger than any previously reported cases.

Case report

A 38-year-old male was referred to our hospital for a large mass in the left thoracic cavity. He presented with cough, sputum, polypnea, and chest congestion after activities for one year. Vascular murmurs were detected below the xiphoid process upon physical examination. Laboratory workup showed elevated levels of NT-ProBNP. Over 20 years ago, a heart murmur was noted but not further investigated.

Coronary computed tomography angiography (CTA) revealed a giant coronary aneurysm measuring 110 × 150 × 217 mm, originating from the left main coronary artery and the left circumflex artery (Fig. 1a,b). Venous phase chest CT identified mural thrombosis in the aneurysm, and a direct communication from the aneurysm to the right ventricle via a dilated and tortuous fistula extending through the right ventricular myocardium (Fig. 1c). The 3D volume-rendering reconstruction highlighted the dilated left coronary sinus and dilated left coronary artery with a giant coronary aneurysm starting at the initial part of the left main coronary artery (Fig. 1d). Left coronary angiography in anterior posterior projection revealed limited development of the aneurysm and the distal portion of the left coronary artery, attributed to the aneurysm's immense size (Fig. 1e). Right coronary angiography in left anterior oblique demonstrated that the left anterior descending coronary artery received retrograde supply from the right coronary artery, indicating occlusion of the proximal portion of the left anterior descending coronary artery (Fig. 1f).

A standard median sternotomy was executed under general anaesthesia with cardiopulmonary bypass following comprehensive preoperative preparations. Cardiac arrest was induced by antegrade cardioplegia through the right coronary artery. A giant coronary aneurysm was observed to arise from the left coronary artery and drainage into the right ventricle via a tortuous and dilated fistula. The aneurysm was incised and the thrombus removed. The entrance hole, communicating directly to the left coronary sinus, and the exit hole, connecting with the distal fistulous tract to the right ventricle, were both sutured, and the former was blocked by a biological mesh for cardiothoracic surgery. A portion of the aneurysm wall was resected, and the residual wall was sutured. A 5 mm incision was created at the distal end of the left anterior descending coronary

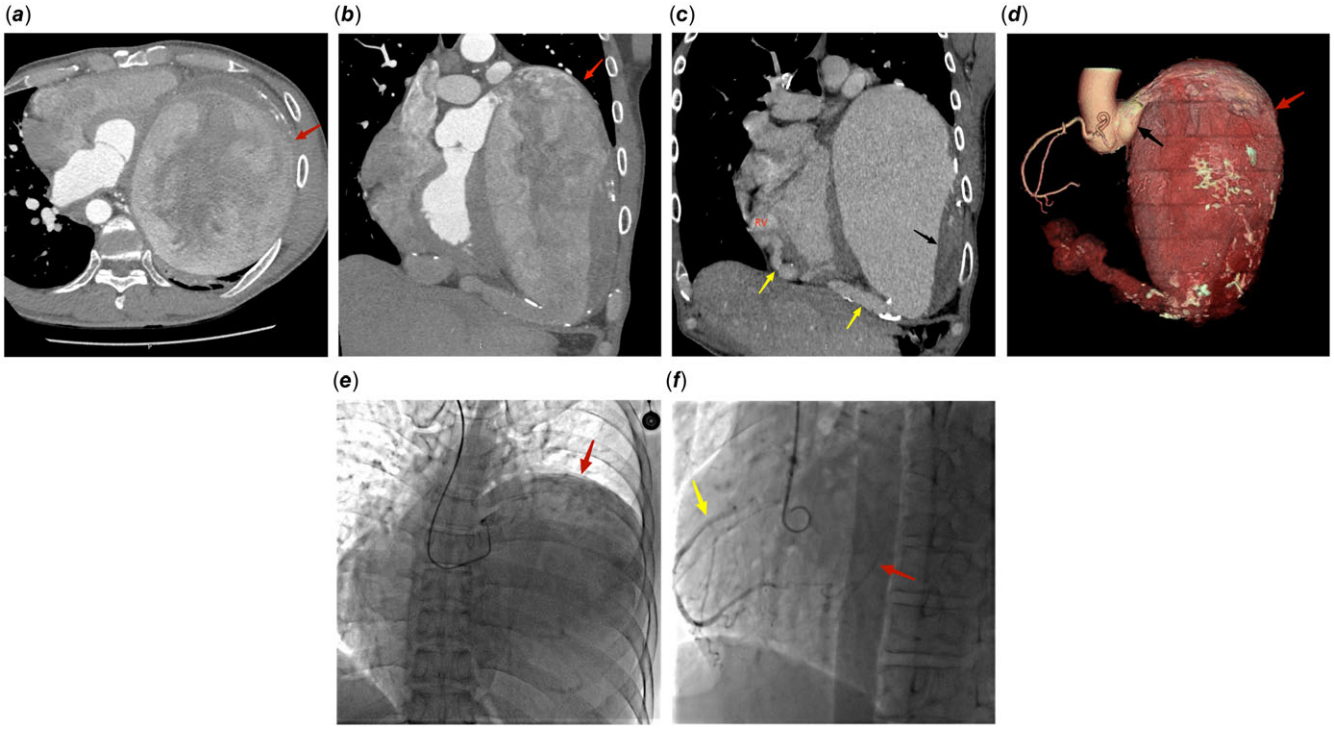

Figure 1. ***a***,***b*** Coronary CTA showed a giant coronary aneurysm (GCAA) (red arrows). ***c*** Venous phase chest CT identified mural thrombosis (black arrows) in the aneurysm and a dilated and tortuous fistula (yellow arrows). ***d*** The 3D volume-rendering reconstruction highlighted the dilated left coronary sinus (black arrows) and dilated left coronary artery with a GCAA (red arrows). ***e*** Left coronary angiography in the anterior posterior projection revealed limited development of the aneurysm (red arrows) and the distal portion of the left coronary artery, attributed to the aneurysm's immense size. ***f*** Right coronary angiography in the left anterior oblique projection demonstrated that the left anterior descending coronary artery (red arrows) received retrograde supply from the right coronary artery (yellow arrows).

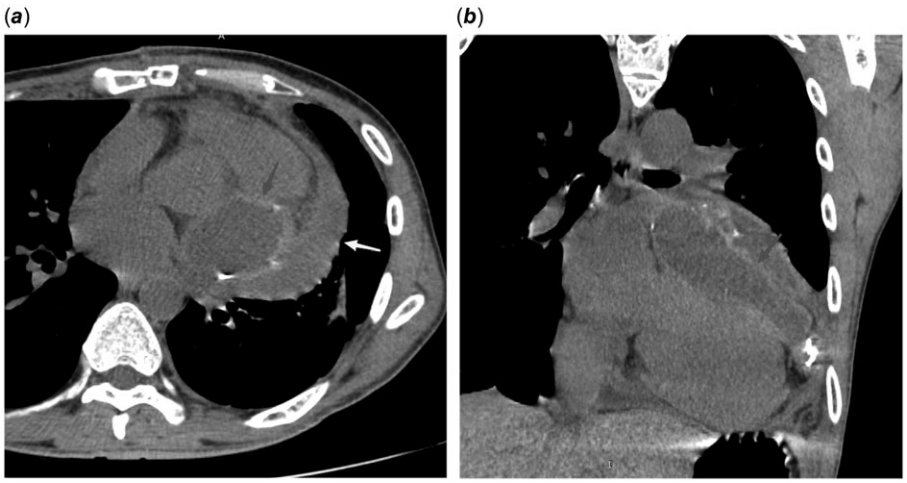

Figure 2. ***a***,***b*** Postoperative chest CT revealed a residual coronary aneurysm wall adjacent to the left ventricle and pericardial effusion.

artery, which was then examined with a bougie to ensure it was unobstructed. An anastomosis was established between the left anterior descending coronary artery and the left internal thoracic artery. Postoperative chest CT revealed a residual coronary aneurysm wall adjacent to the left ventricle and pericardial effusion (Fig. 2a,b). Following symptomatic treatment, the patient recovered within 2 weeks and was discharged from the hospital.

Discussion

In this article, we presented a rare case of a successful surgical intervention for a giant coronary aneurysm with a right ventricular fistula. The pathogenesis of coronary aneurysm remains unclear, but potential factors such as congenital coronary artery fistulas, atherosclerosis, Takayasu arteritis, Kawasaki disease, connective tissue disease, infections, and iatrogenic complications may contribute to the formation of a giant coronary aneurysm. For our patient, a congenital coronary artery fistula was deemed the most probable underlying cause of the aneurysm. The aberrant coronary flow, characterised by turbulent movement between the systolic and diastolic phases due to the abnormal connection between the left coronary artery and the right ventricle, likely stretched the vessel wall. Additionally, compression of the vasa-vasorum by pulsatile flow might cause intermittent ischaemic damage, leading to partial necrosis or

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degeneration of the outer media.[4] These changes might induce the formation of a giant coronary aneurysm. A giant coronary aneurysm can lead to precipitate critical complications such as dissection, rupture, thrombosis, and myocardial infarction.[5] Thus, precise diagnosis and assessment are imperative. In this case, coronary CTA efficiently delineated the origin, fistulous tracts, and drainage site of coronary artery fistula and comprehensive details of coronary aneurysm, increasingly recognised as the imaging method of choice for coronary artery diseases.[6] Coronary angiography contributed further details. Notably, in this case, coronary angiography revealed that the left anterior descending coronary artery received retrograde supply from the right coronary artery, suggesting the aneurysm involved the left main coronary artery and the left circumflex coronary artery, with the proximal segment of the left anterior descending coronary artery occluded.

A standardised treatment protocol for coronary aneurysms remains unestablished. Earlier research highlighted intervention in asymptomatic patients with coronary aneurysm was rare for its poor prognosis. Surgical intervention is deemed suitable for complicated coronary aneurysms at a high risk of rupture, affecting significant bifurcations, and symptomatic patients not eligible for percutaneous coronary intervention.[7] In this case, due to the elevated risk of rupture and the potential for thrombus detachment, surgical management was imperative. Furthermore, the surgical suturing of the orifice at the level of the left coronary sinus led to the closure of the proximal segment of the left coronary artery; hence, it was crucial to reestablish the blood flow to the left anterior descending coronary artery using coronary artery bypass grafting. The results affirmed the success of surgical management for this giant coronary aneurysm.

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Competing interests. None.

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