CASE REPORT

Right coronary artery with multiple giant aneurysms fistulizing into left ventricle

Jing Lu[1], Heshui Shi[2]

[1]*Department of Nuclear Medicine, ZhongShan Hospital Xiamen University, Xiamen, 361000, China*
[2]*Department of Radiology, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, Wuhan, 430022, China*
Correspondence: Heshui Shi
E-mail: heshuishi@hotmail.com
Received: May 25, 2016
Published online: July 05, 2016

To cite this article: Jing Lu *et al*. Right coronary artery with multiple giant aneurysms fistulizing into left ventricle. Receptor Clin Invest 2016; 3: e1361. doi: 10.14800/rci.1361.

Introduction

Coronary artery aneurysm (CAA) is an uncommon disease with the incidence of between 1.5% and 5% [1, 2]. In this report a very rare case of CAA will be presented: multiple giant coronary artery aneurysms and coronary ectasia associated with a fistula between right coronary artery (RCA) and left ventricle (LV).

Case report

A 51-year-old male patient was admitted for cough and expectoration. Cough occurred during sleeping, and the condition lasted for half a month. On general examination, pulse rate was 85 per minute and blood pressure was normal. Physical examination showed normal heart sounds and a continuous grade III/VI systolic murmur could be heard in cardiac apex.

Echocardiography showed dilation of RCA with flow to LV (Fig 1). An initial diagnosis of a RCA-LV fistula with aneurysms was made.

In coronary angiography, cloudy image rather than normal sharp of RCA was showed after the injection of contrast medium (Fig 2), combined with the result of echocardiography, which indicated the possibility of aneurysm.

Coronary computed tomography angiography (CCTA) was performed to further identify the abnormality, which revealed the enlargement of the opening in approximately 3cm and diffusely dilated of RCA. The distribution of RCA could be showed graphically by the volume rendering technique (VRT) (Fig 3), which was travelling tortuously into the LV. A saccular aneurysm was found in size of about 12cm × 12cm and located in its proximal portion (Fig 4). Another giant fusiform aneurysm could be found next to the fistula. The fistula was about 2cm in diameter. Right atrium and ventricle were crushed obviously. Pericardial effusion was observed. The LCA was normal.

Under these conditions, the patient underwent open heart surgery for resection of the aneurysm and coronary bypass surgery through median sternotomy. During the surgery, a saccular aneurysm measured approximately 12 cm × 12 cm was found in the proximal portion of RCA, which severely compressed the right atrium and ventricle (Fig 5).

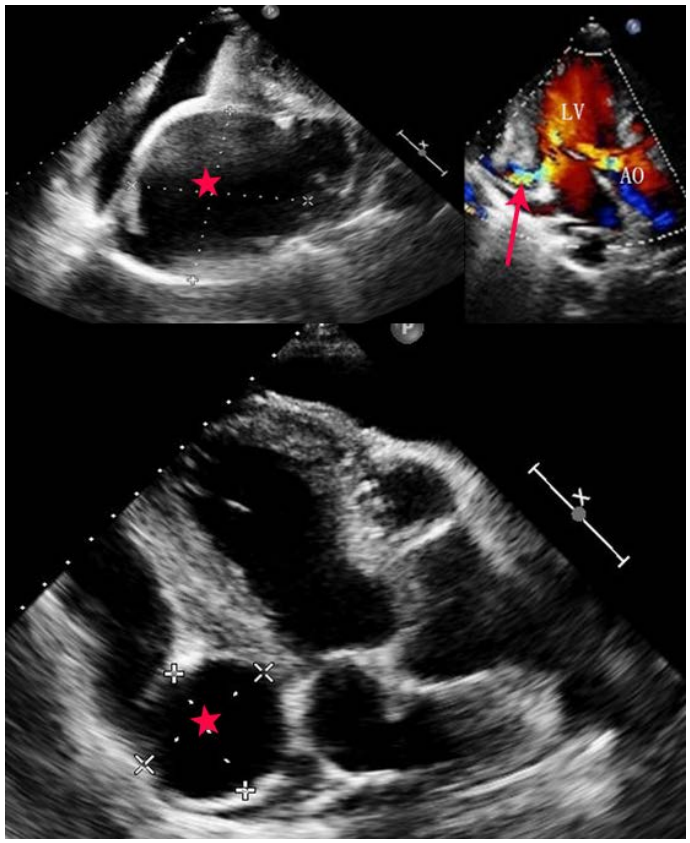

Figure 1. Echocardiography showed aneurysms (star) of RCA with flow to LV (arrow).

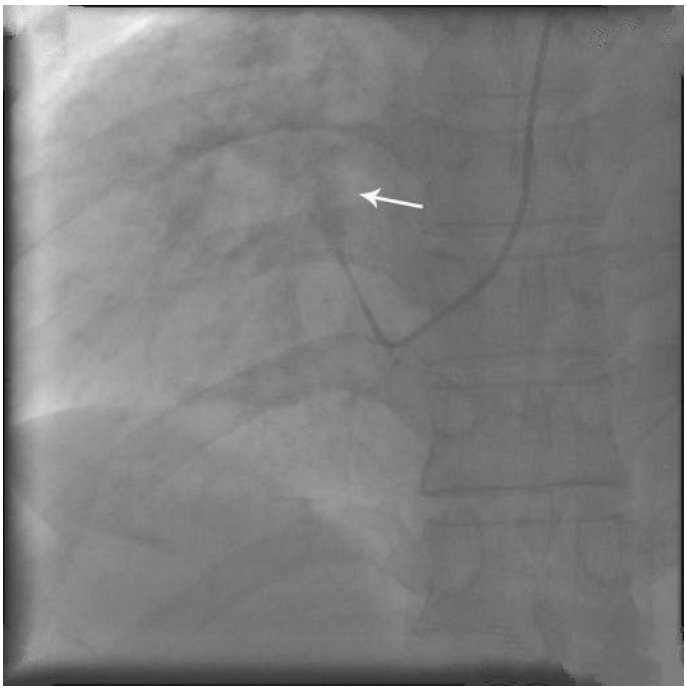

Figure 2. The selective coronary angiography showed the RCA was cloudy after injecting contrast medium (arrow).

Specimen was sent for histopathology. The fistula was located in the opening of left posterior artery. Coronary artery ligation at its origin and fistula were performed for indirect closure of the shunt. A saphenous vein was grafted to reestablish the continuity of the RCA.

Figure 3. VRT showed visually the coronary artery aneurysms (star) and coronary ectasia with fistula to the left ventricle (arrow).

The pathological result showed that the wall was thickened and local mucoid degeneration was found in the vessel media. A large number of red blood cells were observed in the internal elastic lamina (Fig 6).

The patient recovered without complications from the surgery.

Discussion

CAA is defined as the dilatation of coronary artery exceeding the diameter of normal adjacent artery segments or the diameter of the patient's largest coronary artery by 1.5 times [1]. Giant CAA is defined as greater than 2 cm in diameter [3]. This case was diagnosed with giant coronary artery aneurysms and coronary ectasia because aneurysmis restricted to a localized dilatation of coronary arteries which can be saccular or fusiform in shape, and ectasiaisto describe diffuse dilatation which involves the majority of the length of the artery (at least 50%) [1].

In adults, CAA is predominantly atherosclerotic in origin, other causes include Kawasaki disease, Takayasu's arteritis, trauma, infection, idiopathic origin of coronary fistula, congenital malformation and angioplasty [4-6]. Although coronary artery fistulae is infrequent congenital anomalies and only in very rare instances is termination in a left heart chamber, in this case the RCA aneurysm and coronary ectasia were considered resulting from the congenital RCA-to-LV fistula, as the patient did not have a past history of

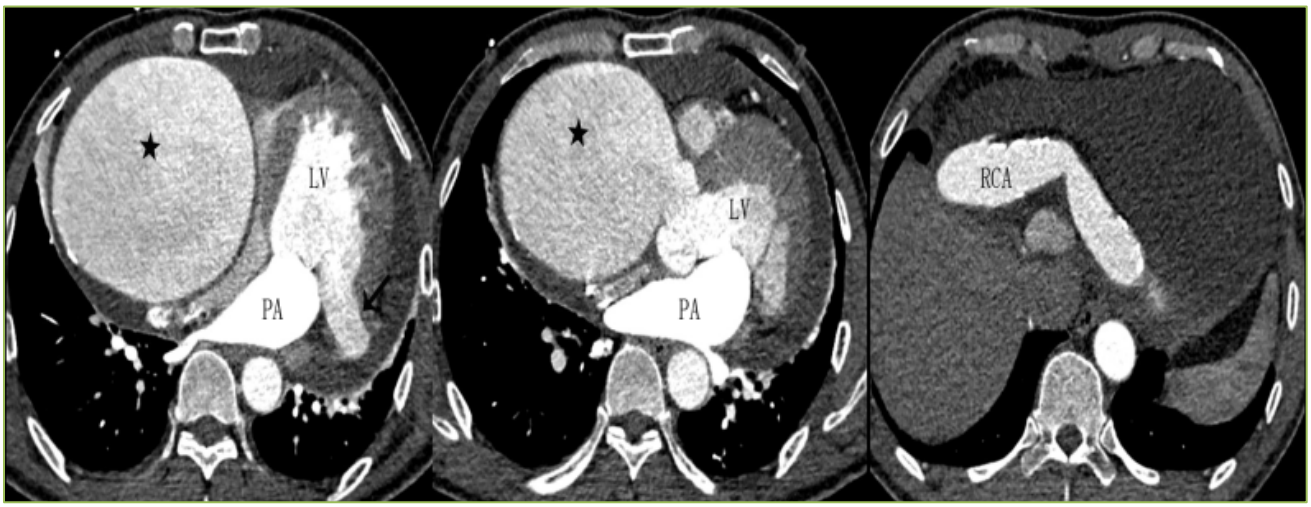

Figure 4. Axial images from CCTA. The opening of the RCA enlarged. There was a giant aneurysm about 12×12cm in dimension and located in its proximal portion (star). The RCA flowed into the left ventricle (arrow).

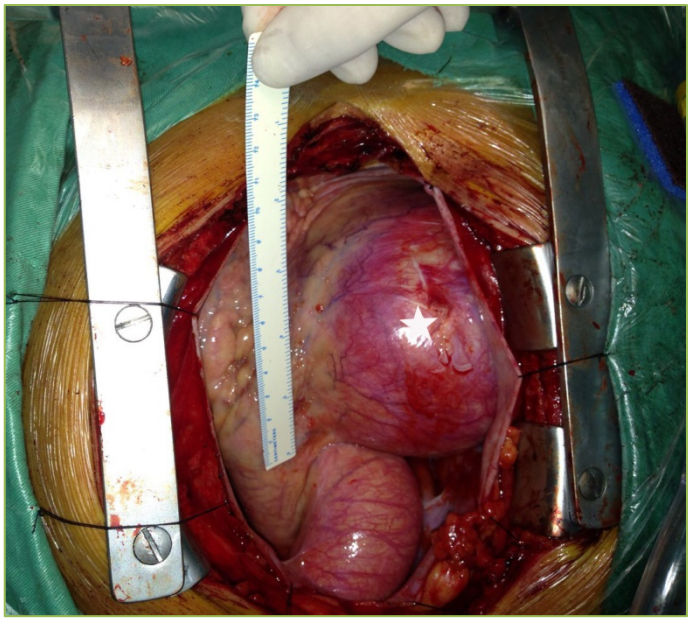

Figure 5. Perioperative view of the heart. A saccular aneurysm measuring approximately 12 cm by 12 cm was found (star).

hypertension or atherosclerosis, or other diseases mentioned above [2,7]. What the microscopic examination of excision revealed neither did not imply distinct etiology such as congenital malformation as the cause.

It was an unusual case as there were multiple giant aneurysms in different positions. It was quite different from other cases in literatures that single aneurysm was usually reported in the vicinity of the fistula. The RCA flow was turbulent to-and-fro between the systolic and diastolic phases due to the fistula. And it could overstretch the vessel wall, and could lead to dysplastic change of the media in the RCA, which was reported to be the pathogenesis of CAA [1, 3, 5]. Besides, the high flow due to the enlarged opening of the RCA may contribute to the proximal aneurysm. And the high resistance from the LV may be part of the causation of wall thickening and the distalis aneurysm. Huge shunting through the big fistula, which could be observed through echocardiogram, could also predispose coronary aneurysm formation by increasing the blood flow in the coronary artery [8]. Pericardial effusion was considered to be transudate due to the giant aneurysms, as there were no infections.

So the final diagnosis of this patient was congenital heart disease of RCA-to-LV fistula with giant right coronary artery aneurysms and coronary ectasia.

CAA may lead to thrombus formation, occlusion, embolization, myocardial ischemia, or myocardial infarction [3]. In this case, the pathological result indicated the thrombus in the largest saccular aneurysm wall without any clinical manifestations (Fig 6).

Conventional coronary angiography is the golden standard of cardiovascular and cerebrovascular diseases. While in this case, the aneurysm in the proximal portion was too large, and contrast medium was diluent and washed away immediately after injection, and the abnormalities could not be observed at all. On the other hand, echocardiography is the initial method for heart diseases, and it discovered aneurysms and fistula exactly in this case. But it could not afford entire vessels with abnormalities visually for doctors and was not sufficient for surgical treatment. Compared to above examination methods, CTA was a more proper method as it not only provided the exact drainage pathway with easy and reliable non-invasive imaging technique, but also provided far better spatial resolution than any other noninvasive

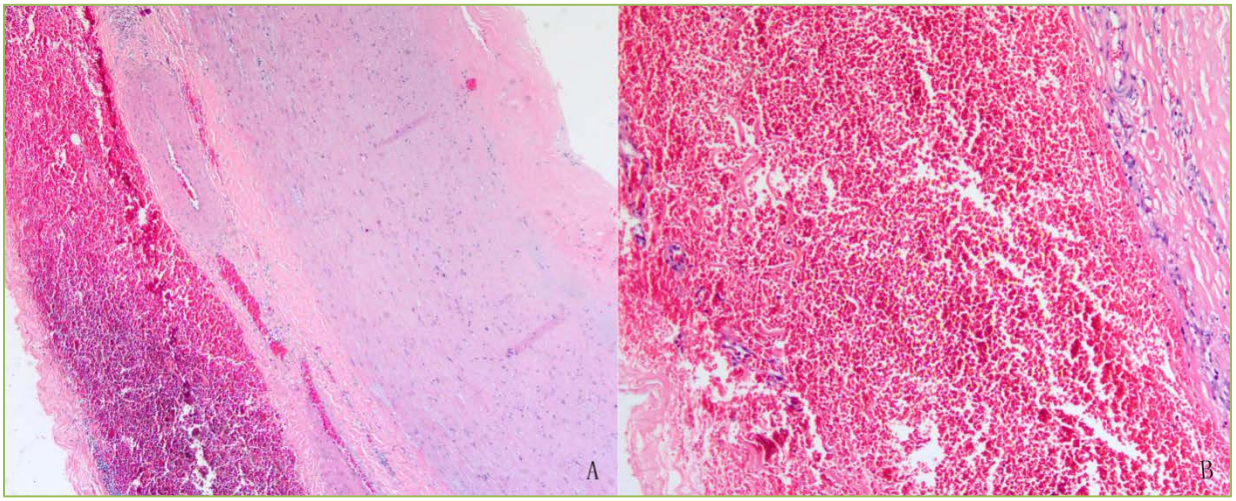

Figure 6. The pathological result indicated that the wall was thickened and local mucoid degeneration was found in the vessel media（A, ×40）. A large number of red blood cells were observed in the internal elastic lamina which was supposed to be thrombus (B, ×100).

imaging modality and allows volumetric reconstruction as well [7].

The appropriate therapy is unknown, and published guidelines have been based on anecdotal experience rather than controlled trials [1]. But surgery has been recommended in general.

Conflicting interests

The authors have declared that no conflict of interests exist.

Author contributions

Jing Lu analyzed this case and wrote this article. Heshui Shi guided and modified the article.

Abbreviations

CAA: Coronary Artery Aneurysm; RCA: Right Coronary Artery; LV: Left Ventricle; CCTA: Coronary Computed Tomography Angiography; VRT: Volume Rendering Technique.

References

1. Syed M, Lesch M. Coronary artery aneurysm: a review. Prog Cardiovasc Dis 1997; 40:77-84.
2. Papadopoulos DP, Ekonomou CK, Margos P, Moyssakis I, Anagnostopoulou S, Benos I, *et al*. Coronary artery aneurysms and coronary artery fistula as a cause of angina pectoris. Clin Anat 2005; 18:77-78.
3. Nichols L, Lagana S, Parwani A. Coronary artery aneurysm: a review and hypothesis regarding etiology. Arch Pathol Lab Med 2008; 132:823-828.
4. Jha NK, Ouda HZ, Khan JA, Eising GP, Augustin N. Giant right coronary artery aneurysm-case report and literature review. J Cardiothorac Surg 2009; 4:18.
5. Liu L, Wei X, Yan J, Pan T. Successful correction of congenital giant right coronary artery aneurysm with fistula to left ventricle. Interact Cardiovasc ThoracSurg 2011; 12:639-641.
6. Burgazli KM, Atmaca N, Mericliler M, Cetin SM, Evagelopoulos N, Erdogan A, *et al*. Giant aneurysm of the right coronary artery with fistula to right atrium. Eur Rev Med Pharmacol Sci 2013; 17:2457-2459.
7. Wei J, Shu Y, Wei X, Tang J, Yan J, Zeng H, *et al*. Right coronary artery fistula to left ventricle complicated with huge coronary artery aneurysm. Intern Med 2011; 50:239-242.
8. Ipek G, Omeroglu SN, Goksedef D, Balkanay OO, Basar I, Ayan F. Giant right coronary artery aneurysm associated with coronary-cameral fistula. Tex Heart Inst J 2012; 39:442-443.