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# A Rare Case of Coronary Artery Fistula Associated with Giant Coronary Artery Aneurysm and Aortic Regurgitation

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**Abstract:** Coronary artery fistula, congenital or acquired, is clinically rare. Congenital Coronary artery fistula is reported incidence of 0.1–0.2% in the adult population referred for cardiac catheterization. Coronary artery fistula with aneurysm is an infrequent vascular anomaly. Angiography and echocardiography have traditionally been used as diagnostic tools for coronary artery fistulas. However, the anatomy of the Coronary artery fistula was then determined in detail by 64-multislice computed tomography angiography. We report a rare case of 54-year-old male patient who had a coronary artery fistula associated with giant coronary artery aneurysm and aortic regurgitation.
**Keywords:** Coronary artery fistula; coronary artery aneurysm; aortic regurgitation.

## INTRODUCTION

Coronary artery fistula (CAF) is the unusual anomalous communications between a coronary artery and a cardiac chamber or major vessel (vena cava, pulmonary veins, pulmonary artery). CAF arises from the right coronary artery (RCA) in approximately half of all patients. CAF draining into the left ventricle that is an extremely rare condition with a reported incidence of 1.2% of all CAF [1]. CAF with giant coronary artery aneurysm (CAA) is very rare. This report describes CAF associated with giant CAA and aortic regurgitation.

## CASE REPORT

A 54-year-old man presented with complaints of exercise-related palpitation and also chest discomfort for two years. He had no specific past medical history. The patient was in New York Heart Association Class III. On physical examination, there was a continuous murmur in the 3rd and 4rd rib of left border of sternum; and a moderate diastolic murmur in the second auscultation area of aortic valve. He had blood pressure of 141/50 mm Hg. The other findings were normal. The chest X-ray showed a cardiothoracic ratio of 75% (Fig.1). Transthoracic echocardiography demonstrated dilation of the left ventricle (left ventricular dimension in end-diastole/end-systole: 86/43 mm), aneurysmal dilatation of the proximal right coronary artery is 55mm (Fig.2A), the distal contorts tortuous, and CAF draining into the left ventricle(LV) (Fig.2B), the diameter is 11.7mm (Fig.2C); and aortic regurgitation (Fig.2D). Selective coronary angiography (CA) demonstrated a right CAA, and tortuous right CAF draining into the LV (Fig.3A), the diameter is approximately 75.7mm (Fig.3B); the other information is unknown. 64-multislice computed tomography angiography (64-MSCTA) showed the maximum diameter of 73mm of aneurysmal dilatation of the proximal right coronary artery, and drainage into the LV, the diameter is 15mm (Fig.4). During the operation, the patient was found to have aneurysmal dilatation and tortuous right coronary artery with a fistula arising from the distal to posterior descending artery origin and draining into the LV immediately inferior to the posterior mitral leaflet (Fig.5, A and B). Aortic annulus was expanded, and right coronary artery valve leaflet was relaxation. The surgical operation of patient included right coronary artery angioplasty, ligation of CAF and replacement of aortic valve. The examination of postoperative 64-MSCTA showed artery pristine and collateral circulation opens (Fig.6). The patient's postoperative course was unremarkable. In the follow-up for five years, patients had showed no abnormalities.

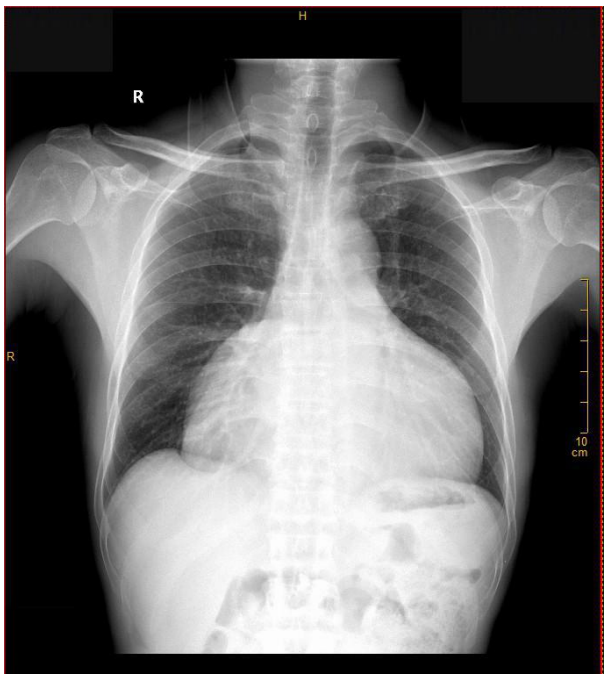


**Fig 1: Chest X-ray showed a cardiothoracic ratio of 75%**

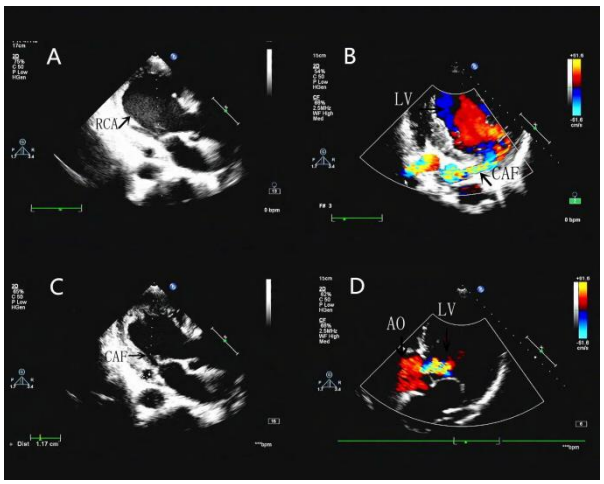


**Fig 2: Transthoracic echocardiography (A): Aneurysmal dilatation of the proximal RCA (arrow) is 55mm. (B): the dilated tortuous RCA (arrow), CAF draining into the LV, (C): the diameter is 11.7mm (arrow) (D): aortic regurgitation**

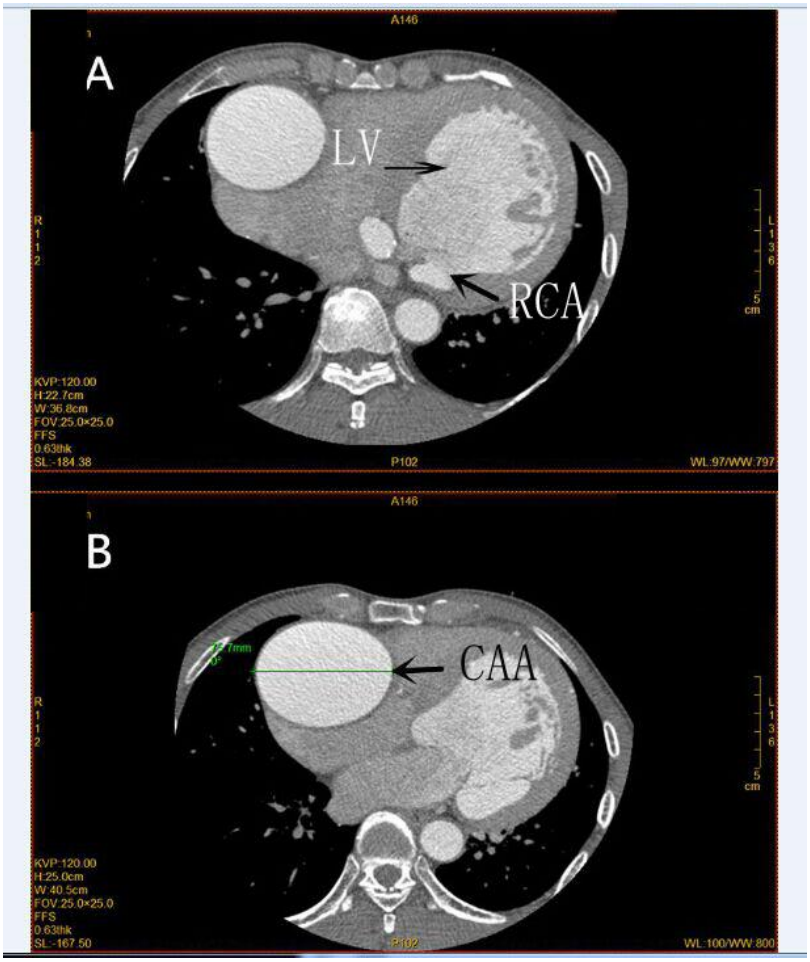


**Fig 3: Axial computed tomography (A): RCA with connection to LV, the arrowhead points the fistula; (B): the maximum diameter of 75.7 mm of CAA (arrow)**

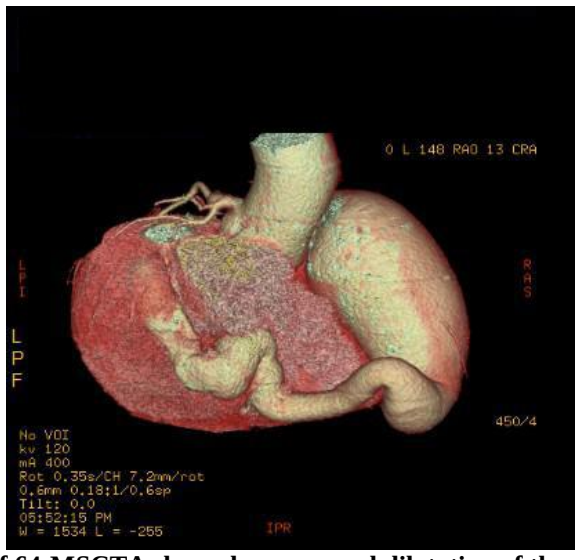


**Fig 4: Reconstruction image of 64-MSCTA showed aneurysmal dilatation of the proximal RCA and tortuous of distal RCA**

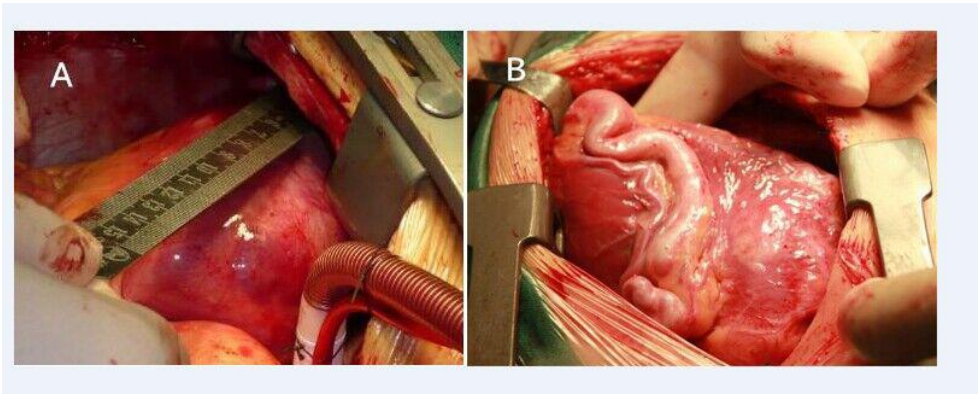


**Fig.5 (A) Operative photograph of aneurysmal dilatation of the proximal RCA. (B) Operative photograph of the dilated tortuous RCA.**

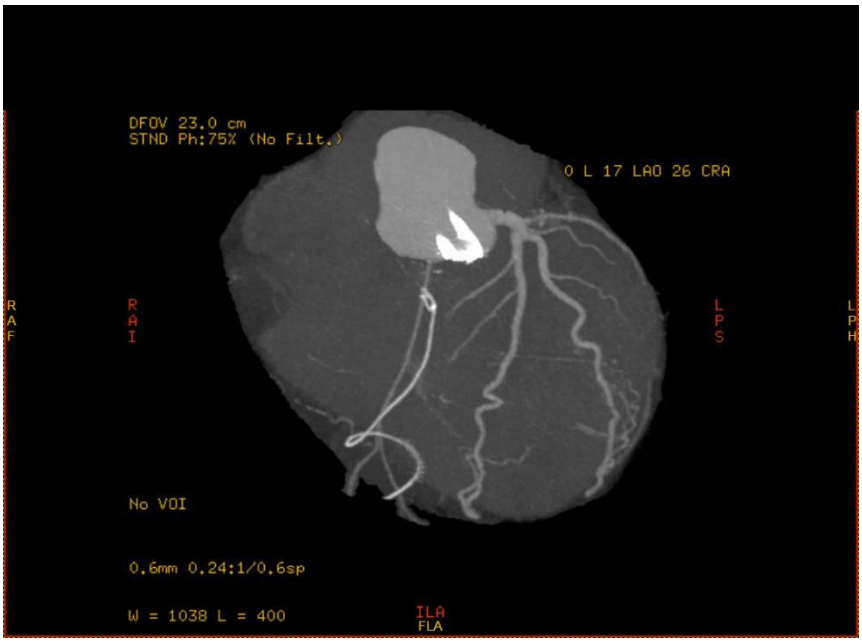


**Fig.6 Postoperative reconstruction of 64-MSCTA image showed unobstructed and established of compensatory circulation of RCA**

## DISCUSSION

CAF is a rare anomaly of the coronary arteries, not differences of race, and discovered at any age. CAF has unilateral, bilateral or multilateral fistulas. The 80% of all patients were unilateral fistulas, 18% of bilateral fistulas and 2% of multilateral fistulas [2]. It was first described by Krause in 1865, and the first surgical treatment was done by Bjork and Crafoord in 1947. It may be congenital or acquired, but acquired CAF has been reported rarely. The latter may occur due to various reasons; for instance, thoracic trauma, Cardiac surgery, acute myocardial infarction, Cardiac catheterization, Angioplasty, Pacemaker implantation, endomyocardial biopsy, and so on.

Approximately half of all patients with CAF do not reveal any symptoms. The symptoms mainly depend on the size of the fistula, on the receiving chamber, myocardial ischemia and occasionally high output congestive heart failure. About 50% of CAF originate from the RCA, 42% from the left coronary artery (LCA) and 5% from both of them. The 41% of total patients with CAF drain into the right ventricle(RV), 26% into the right atrium(RA), 17% into the pulmonary artery(PA), 7% into the coronary sinus(CS), 3% into the LV and 1% into the superior vena cava(SVC) [3,4].

The Symptoms of patients with CAF may present irritability, fatigue, palpitations, and dyspnea on exertion, fatigue, orthopnea, chest pain, endocarditis, arrhythmias, stroke, high-output congestive heart failure, myocardial ischemia, or myocardial infarction. It is presumed that ischemic symptoms are caused by “steal phenomenon” [5].Complications of CAF are secondary aortic valve disease, myocardial ischemia, bacterial endocarditis, congestive heart failure, sudden cardiac death, etc. complications are present in 11% of patients younger than 20 years and in 35% of patients older than 20 years of age [6].

The methods of diagnosis have echocardiography, conventional contrast CA, magnetic resonance (MR) imaging and CTA. Echocardiography can identify large fistulas, although it may reveal high-volume flow by color-flow, left atrial (LA) and LV enlargement, imaging Drainage of the fistula and dilatation of the coronary artery. Although CA is the gold standard diagnostic test for detection of CAF, it can demonstrate the anatomy, size, number, origination and termination site of the fistulas; but its two-dimensional nature, CA cannot show reliably the relationship of aberrant vessels with the underlying cardiac structures, it is clearly invasive and associated with procedural morbidity (1.5%) and mortality (0.15%) risks [7]. However, the spatial resolution of MRI is inadequate for coronary artery imaging, especially for meticulous analysis of the distal arterial course. Moreover, the temporal resolution of MRI is unsatisfactory, it takes a bit longer. CTA is a non-invasive 3D imaging technique that provides an excellent distal coronary artery and can be used for the assessment of complex vascular anatomy by using electrocardiographically gated reconstruction methods, and could be helpful for planning future cardiovascular therapeutic approaches, either interventional or surgical.

Management of CAF is still a controversial issue, especially in asymptomatic patients. CAF with asymptomatic may close spontaneously but is rarely occur in small fistulas. Small fistulous connections in the asymptomatic patient may be treated conservatively. Medical treatment with either calcium channel blockers or beta blockers is suggested. Symptomatic patients with CAF require closure through surgical ligation or percutaneous therapeutic embolization (PTE). The main indications for surgery ligation are high fistula flow, multiple communications and terminations, significant aneurysmal formation, very tortuous pathways, et al [8]. On the other hand, indications for PTE include older patient age, extraanatomic termination of CAF away from the normal coronary arteries, single drain site, proximal location of the fistulous vessel and so on [9]. However, PTE has been widely used in recent years; in addition, PTE and Surgical ligation show that both approaches have similar early effectiveness, morbidity, and mortality.

## CONCLUSION

Coronary artery with a diameter more than 20 mm is termed as "giant aneurysm". The diameter of coronary artery aneurysm of our patient is 75.7mm. It is a very rare case. The treatment methods of CAF choose surgery ligation or PTE, it should be based upon the anatomical and complication and functional characteristics of the fistula. According to the patient individually, we did treatment that the surgical operation included right coronary artery angioplasty, ligation with a running suture of CAF and replacement of aortic valve. The patient’s postoperative course was unremarkable. Therefore, surgical treatment CAF is a safe and effective method.

**CONFLICT OF INTERESTS:** None declared.


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