

## Surgical Treatment of Coronary Artery Aneurysm with Coronary Artery Fistula

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**Coronary artery aneurysm (CAA) is an uncommon disease with an incidence of 1%–5% in evaluated patients. Atherosclerosis is its most common etiology in adults, occurring in 50% of cases. Also, CAA is associated with predisposing factors such as Kawasaki disease, stent-angioplasty, and notably, coronary artery fistula. Most CAAs are asymptomatic but occasionally lead to life-threatening conditions, such as rupture, infection, and myocardial ischemia. Thus caution is urged for cardiac surgeons. We here report on seven surgical cases of CAA with coronary artery fistula in our institute, focusing on their surgical treatment. (Ann Thorac Cardiovasc Surg 2009; 15: 198–202)**

**Key words:** coronary artery aneurysm, coronary artery fistula, surgical treatment

### Introduction

Because of the spread of multidetector-row computed tomography (MDCT) or coronary angiography (CAG), the numbers of coronary artery aneurysms (CAAs) have increased recently. CAA is defined as dilatation that is more than 1.5 times the diameter of normal adjacent segments.<sup>1)</sup> Kawasaki disease (mucocutaneous lymph node syndrome) is well known for affecting the coronary artery, resulting in aneurysmal change and rupture.<sup>2)</sup> Half of all CAAs in adults are caused by atherosclerosis.<sup>1)</sup> Among them, CAAs with coronary artery fistula (CAAAF) is a noteworthy condition because shunt predisposes fistulous vessels to frequent aneurysmal change and enlargement leading to rupture. Therefore cardiovas-

cular surgeons must be alert to its etiology, characteristics, and countermeasures.

### Case Reports

From 2000 to 2008, we encountered seven surgical cases of CAAAF (Table 1), representing 0.73% of the patients undergoing isolated coronary artery bypass grafting in the same period. Mean age was  $65.4 \pm 18.3$ , and all patients were female. Three patients were symptomatic with dyspnea, but it was unclear whether it stemmed from left-to-right shunting. In asymptomatic patients, an abnormal cardiac shadow on a chest radiograph (case 1/7), deep-vein thrombosis (case 3), and coagulation disorder (case 6) were the reasons for further investigation and detection of CAAAF.

All patients underwent CAG, and CAAAFs were identified preoperatively. As for fistula types, shunts from the left anterior descending branch (LAD) to the pulmonary trunk (PA) accounted for 85.7% of cases (6), from the right coronary artery (RCA) to the PA for 42.8% of cases (3), and from the RCA to the right ventricle (RV) for 14.2% of cases (1). The maximal diameter of the CAA in each patient was in the range of 11.5–80 mm (mean  $40.9 \pm 23.6$  mm), and there were no ruptures. Cases 1/2 had isolated significant coronary

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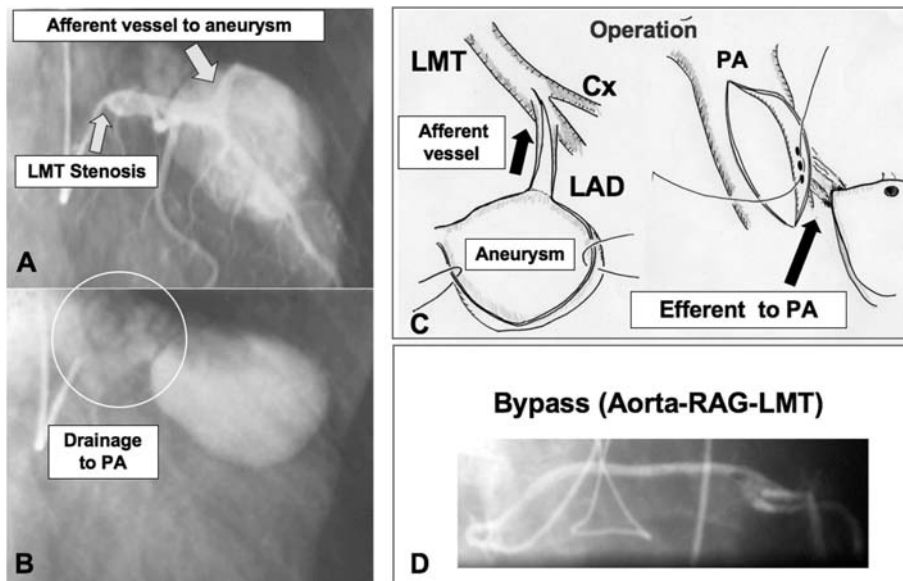
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**Table 1. Summary of seven surgical cases of CAAAF in our hospital**

Age and sex	Symptom	Fistula	Maximum diameter (mm)	Coronary stenosis	Shunt ratio
1. 77 F	(-)	LAD → PA	50	LMT: 90%	20%
2. 72 F	Dyspnea	LAD → PA	11.5	#7: 70%; #9: 50%; #11: 60%	Unknown
3. 77 F	(-)	LAD → PA	14	(-)	13.6%
4. 30 F	Dyspnea	RCA → RV	38	(-)	46%
5. 75 F	Dyspnea	RCA/LCA → PA	80	LAD total?	Unknown
6. 61 F	(-)	LAD → PA	40	(-)	26%
				Anterior ischemia	
7. 66 F	(-)	RCA/LAD → PA	53	(-)	43%

All patients were females, and the maximal diameter was 80 mm in Case 5.

F, female; LAD, left anterior descending branch; PA, pulmonary trunk; RCA, right coronary artery; RV, right ventricle; LCA, left coronary artery; LMT, left main trunk.



**Fig. 1.** Case 1 is presented.

**A,B:** Preoperative left coronary angiographs demonstrate a large CAA with the fistula from the LAD to the PA.

**C:** Perioperatively, the PA and CAA were incised and the fistulae were closed.

**D:** Aortocoronary bypass grafting with radial artery graft was added because of LMT stenosis.

LMT, left main trunk; PA, pulmonary artery; Cx, circumflex artery; LAD, left anterior descending branch; RAG, radial artery graft.

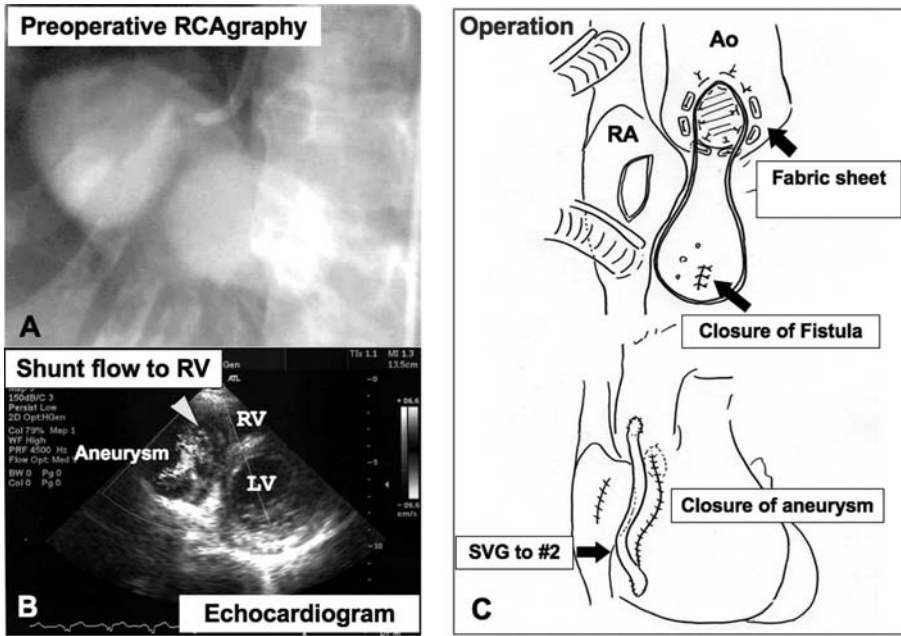
stenoses. Meanwhile, the LAD of case 5 was not identified on CAG preoperatively because of compression by the large aneurysm. Myocardial scintigraphy in case 6 demonstrated anterior hypoperfusion. However, there was no significant stenosis on CAG, indicating the presence of coronary flow steal through the fistula. A high-grade left-to-right shunt reaching 40% was seen in case 4 and case 7. Under cardiopulmonary bypass (CPB) and cardiac arrest, all aneurysms were incised, and fistulae were basically closed with sutures from the inside. Coronary bypass grafting was performed if necessary. All patients recovered well with no complications or recurrence. Three representative cases are described below.

### Case 1

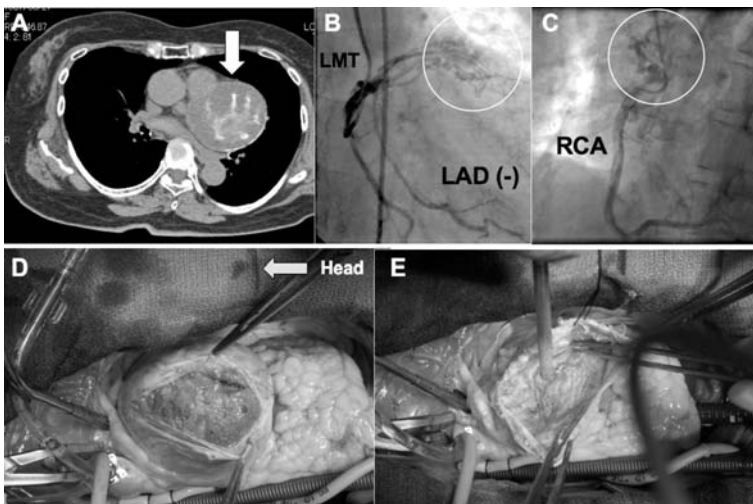
Preoperative CAG showed a large aneurysm (50 mm) with the shunt from the LAD to the PA (Figs. 1A and 1B). After cardiac arrest, the aneurysm was incised to identify the afferent and efferent vessels from the inside. The afferent vessel was traced back to the LAD. The PA was incised, and efferent vessels were identified and closed with sutures (Fig. 1C). The radial artery graft was sewn into the incised proximal LAD, and an aortocoronary (AC) bypass was added (Fig. 1D).

### Case 2

The RCA aneurysm (Figs. 2A and 2B) was longitudinally incised, and the enlarged RCA orifice was identified. The right atrium (RA) was opened small, and a car-



**Fig. 2.** Case 4 is presented. RCA angiogram (A) and echocardiogram (B) demonstrate the giant RCA aneurysm with a shunt to the RV. Perioperatively, the aneurysm was incised longitudinally and fistulae were identified with the injection of cardioplegic solution from the RV. The enlarged RCA orifice was closed with the use of a fabric sheet, and the aneurysmal wall was folded and closed. An AC bypass (SVG-#2) was added (C). RCA, right coronary artery; RV, right ventricle; LV, left ventricle; Ao, aorta; RA, right atrium; SVG, saphenous vein graft.



**Fig. 3.** Case 5 is presented. A: The CT shows a giant aneurysm (8 cm) with calcified hematoma on the left side of the PA (arrow). B,C: The coronary angiographs show nidus vessels from the RCA and LCA to the PA. D: Intraoperative photographs show the incised giant aneurysm filled with hematoma. E: Afferent and drainage vessels of the aneurysm were examined from the inside. LMT, left main trunk; LAD, left anterior descending branch; RCA, right coronary artery.

dioplegic cannula was inserted to the RV through the RA. Blood cardioplegic solution was injected into the RV so that the fistulae could be identified. The RCA orifice was closed with the use of a sheet (Woven Double Velour Fabric, Hemashield, Boston, USA), and the aneurysm was folded and closed (Fig. 2C). The AC bypass to the distal RCA was done with a saphenous vein graft (SVG).

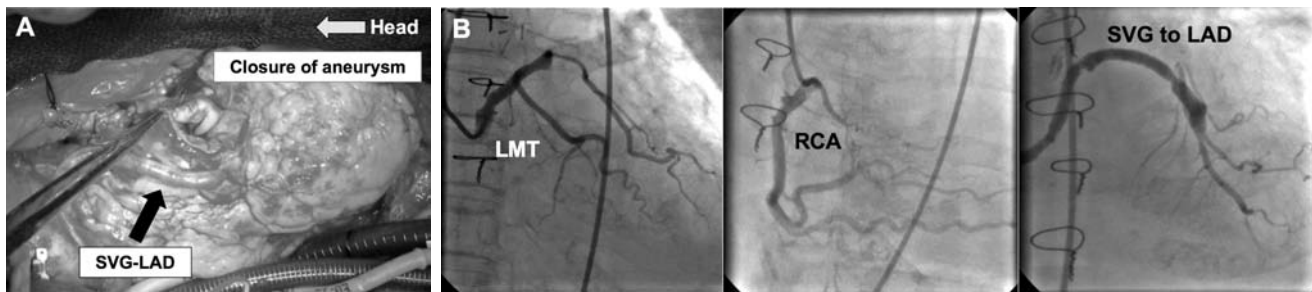
### Case 3

A chest CT demonstrated a giant aneurysm (80 mm) (Fig. 3A), which was preoperatively thought to be a malignant tumor. Preoperative CAGs (Figs. 3B and 3C) show shunt

vessels from the RCA and LAD to the PA. Perioperatively, the aneurysm was found to be filled by a clot (Fig. 3D). Afferent and efferent vessels were examined and closed from the inside. It was shown that the LAD was pushed forward by the CAAAF. An AC bypass to the LAD was performed using an SVG (Fig. 4A). Postoperative CAGs demonstrated no residual shunt or a patent SVG to the LAD (Fig. 4B).

### Discussion

Coronary artery fistula is an uncommon innate anomaly



**Fig. 4.** Case 5 is presented.

**A:** The giant aneurysm is closed.

**B:** Coronary artery bypass grafting (SVG-LAD) was done, and postoperative CAGs demonstrate no residual shunt vessel or a patent SVG to the LAD.

SVG, saphenous vein graft; LAD, left anterior descending branch; LMT, left main trunk; RCA, right coronary artery.

with an incidence of 0.2%–1.5% among patients evaluated by coronary angiographic studies.<sup>3,4)</sup> The coronary artery fistula is characterized by shunts from the coronary artery to the PA, RV, or coronary sinus. The shunt from the LAD branch to the PA seems to be the more common type, which accounts for 40%.<sup>3,4)</sup> Occasional cardiac symptoms such as chest pain or dyspnea can be attributed to the large amount of shunt or coronary flow steal;<sup>5)</sup> care is therefore urged.

Of note, CAAs occur frequently in approximately 19%<sup>6)</sup> of patients with coronary artery fistula. By definition, a fistula consists of a “nidus” of vessels with fragile smooth muscles. Considering the constant exposure to arterial pressure and much blood flow, dilatation and aneurysmal change with age would be a logical outcome. Actually, CAAAFs are common in elderly patients. It is interesting that giant CAAAFs (30 to 40 times the diameter of a normal coronary artery [which is 2–3 mm]) are occasionally reported.<sup>7,8)</sup> We encountered such a case (80 mm), which was preoperatively misdiagnosed as a tumor.

The natural course of CAAAF is obscure. The rupture rate is still unknown, but ruptured cases seem to be very rare.<sup>9)</sup> Thus the surgical indications for CAAAF are somewhat controversial, though it would be a small problem if it were possible to obtain independent confirmation of cardiac pathology, such as significant coronary stenosis or valvular diseases. Because the diameter of ruptured CAAAFs can range from 10 to 50 mm,<sup>10)</sup> a relatively small CAAAF or coronary artery fistula in itself<sup>11,12)</sup> should not be treated lightly. It is obvious that rupture,<sup>9,11–13)</sup> infection,<sup>14)</sup> congestive heart failure, or coronary distal ischemia resulting from coronary flow steal<sup>5)</sup> are surgical indications of CAAAF. Syed and Lesch<sup>1)</sup>

describe that there is no difference in the prognosis of “mere CAA” (atherosclerotic CAA) during a short observation period between patients followed up medically and patients surgically treated. As mentioned, however, CAAAF occurs under factors predisposing to frequent aneurysmal change (fragile smooth muscles, much arterial flow, aging, and atherosclerosis). Even if medical treatment is chosen, strict follow-up is mandatory, and the risk of life-threatening rupture remains. Viewed in this way, surgical treatment is a reasonable option for CAAAF. Without doubt, large, saccular, rapidly growing, or symptom-causing aneurysms are indicators for open-heart surgery. Meanwhile, more than a few successful cases of catheter intervention with implantation of coil or covered stent<sup>15)</sup> have been reported. From the viewpoint of less invasiveness, catheter intervention for the CAAAF draining to the PA is a good countermeasure, especially in patients with low-grade shunt flow, small aneurysms, or high surgical risk being strictly followed up.

Surgical treatment strategies require CAG or MDCT for precise planning. In our institute, aneurysms are incised under CPB and cardiac arrest. Therefore it is possible to identify afferent and efferent vessels from the inside. And we usually incise the PA, which facilitates the exclusion of all fistulous drainage vessels steadily without damaging other structures. The purpose of this surgery is to guarantee that not only aneurysms, but also nidus vessels are excluded. Certainly off-pump surgery has succeeded in some cases,<sup>16)</sup> but it has also failed in some.<sup>12)</sup> The possibility should be considered that a remnant shunt might fail to decompress aneurysms and fistulous vessels and induce rupture or recurrence of aneurysms.

Our practical surgical procedures are shown below: (1) The procedure with the use of CPB and cardiac arrest. (2) Incision of the PA, confirmation of drainage vessels with the injection of cardioplegic solution, and closure. (3) Opening of the aneurysms and exclusion by suturing afferent and efferent vessels. (4) Bypass grafting if necessary. It is of interest that our cases were all females. Accordingly, a previous study reported that female patients are more common than males and have a higher risk of rupture.<sup>17)</sup> Given that the most common cause of CAA is atherosclerosis, it is similarly predicted that most patients with CAAAF are male. But in fact this is not so. Pathological, genetic, or molecular biological studies of CAAAF are expected in the future.

## Conclusion

We reported seven surgical cases of CAAAF. The surgical indications for it are still controversial, but life-threatening complications such as rupture and cardiac tamponade can occasionally arise. Therefore open-heart surgery is a reasonable option and should be considered proactively. For the sure and steady exclusion of CAAAFs and fistulous vessels, the use of CPB with cardiac arrest and PA incision is recommended.

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