Multiple giant coronary artery aneurysms with extended coronary ectasia emerging 12 years after previous coronary artery bypass grafting

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Abstract
A 73-year-old man presented with multiple giant coronary artery aneurysms. Twelve years prior to the presentation, he had undergone coronary artery bypass grafting. At that time, he exhibited small aneurysms (16 mm diameter) in the right coronary artery and a single aneurysm (10 mm diameter) in the left circumflex artery. During follow-up, the aneurysms gradually increased in size (to 45 and 30 mm, respectively, at 12 years after surgery). We resected all of the aneurysms and performed coronary artery bypass grafting of the left circumflex artery through re-sternotomy.

Keywords
Coronary artery aneurysms, coronary artery ectasia, post operation

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Introduction
Coronary artery aneurysms (CAAs) with diameters exceeding 4 cm, defined as “giant,”1,2 are very rare (0.02% incidence).3 The natural history and prognosis of CAAs remain unclear.1–5 Progression may be asymptomatic, but fatal complications may develop.2–5 The treatment options include medical management, percutaneous coronary interventions, and surgery.2–4 However, no consensus on the optimal treatment, timing thereof, or surgical indications has emerged.2,4 Here, we report a case of successful re-operation to treat multiple giant CAAs that had grown over 12 years after initial coronary artery bypass grafting (CABG). Frequent imaging after the initial CABG revealed the details of aneurysm progression.

Case report
A 73-year-old man presented with multiple CAAs. Twelve years prior to the presentation, he had undergone CABG at another hospital. He also had undergone aortic replacement of an abdominal aortic aneurysm at the age of 63 years and thoracic endovascular aortic repair of a thoracic aortic aneurysm at the age of 69 years. The CABG was performed as the left internal thoracic artery to the left anterior descending artery, radial artery to the first diagonal branch and a saphenous vein graft to the posterior descending artery (Figure 1(a)). At the time of this operation, the maximum diameter of the CAAs in the right coronary artery (RCA) was 16 mm (Figure 1(b)), and that of the aneurysm in the left circumflex artery (LCX) was 10 mm (Figure 1(c)). Regularly scheduled postoperative follow-up imaging revealed gradual growth of all of the CAAs; those in the RCA and LCX attained diameters of 29 and 19 mm, respectively, at 8 years and 45 and 30 mm, respectively, at 12 years after CABG. The images were obtained in the hospital in which the CABG was performed, but the surgery had been considered to be a high-risk option due to the patient’s comorbidities. As soon as the patient was referred to us, we scheduled surgery. He was
asymptomatic and had no history of myocardial infarction. He was given an anticoagulant to treat atrial fibrillation. Preoperative coronary angiography and enhanced computed tomography revealed three CAAs (45, 28, and 26 mm in diameter) in the RCA and one CAA (30 mm in diameter) in the LCX (Figure 2). The distal regions of the right CAAs and
Intraoperative photographs of the (a) CAAs in the right coronary artery. (b) The right CAAs were opened, and the thrombi were removed. The arrow shows the proximal ostium. (c) The CAA in the left circumflex artery. Note that the intraoperative photographs reflect the surgeon’s view; the left side is cranial and the right side is the caudal. (d) Three-dimensionally reconstructed computed tomographic images show that the CAAs disappeared after the second surgery.

Figure 3. Intraoperative photographs of the (a) CAAs in the right coronary artery. (b) The right CAAs were opened, and the thrombi were removed. The arrow shows the proximal ostium. (c) The CAA in the left circumflex artery. Note that the intraoperative photographs reflect the surgeon’s view; the left side is cranial and the right side is the caudal. (d) Three-dimensionally reconstructed computed tomographic images show that the CAAs disappeared after the second surgery.

Discussion

A CAA is defined as a focal dilation of the coronary artery that is 1.5-fold greater than the adjacent segments;\textsuperscript{2–7} a giant CAA exceeds 4 cm in diameter.\textsuperscript{1,2} Li et al.\textsuperscript{3} reported that the incidence of giant CAAs in the cardiac surgical population was only 0.02%. Coronary artery ectasia (CAE) is the diffuse dilatation of ≥50% of the coronary artery.\textsuperscript{6} Markis et al.\textsuperscript{8} classified CAE topographically into four types with potential prognostic implications; patients with types I and II CAE experience the worst outcomes. Our patient had diffuse type-I CAE of the RCA and LCX.

The etiology of CAA is variable, and includes atherosclerosis, Kawasaki disease, vasculitis, trauma, autoimmune diseases, and connective tissue diseases.\textsuperscript{2–6} Recent reports have described CAAs associated with immunoglobulin G4-related disease.\textsuperscript{9} Our patient had no history of Kawasaki disease or percutaneous coronary intervention. Given the multiple aneurysms in the aorta, we considered immunoglobulin G4-related disease.
disease, but the patient’s serum immunoglobulin G4 level was low. Based on these findings and the pathological results, atherosclerosis conceivably caused the CAAs.

Although the complications of a CAA, including thrombosis, distal embolization, compression of adjacent structures, and rupture, are life threatening,2–4 no consensus has emerged in terms of CAA management.2–4 The treatment options include medical management, percutaneous coronary interventions, and surgery.2–4 In our case, follow-up imaging was done annually in the hospital in which the initial CABG was performed. The anticoagulant given to treat atrial fibrillation may have protected against embolization. Given the rarity of the condition and the very invasive nature of the surgical option, treatment and its timing should be tailored to the patient’s age, comorbidity status, and frailty.

Several surgical indications for CAA have been proposed; they include CAA accompanied by coronary artery stenosis5 and CAA >30 mm.10 The gradual aneurysmal growth (beyond the extent associated with a risk of rupture) was the decisive factor in our selection of surgery. The surgical treatment of giant CAAs usually involves suturing of the afferent and efferent vessels and (if necessary) bypass grafting.3,4 The available information on initial graft patency greatly influenced our surgical plan. The RCA was not involved in our CABG because the initial graft was occluded with collateral flow; our CABG addressed only the LCX.

**Conclusion**

We present a rare case of multiple giant CAAs with extended CAE in a 73-year-old man with multiple cardiovascular comorbidities; he underwent re-operation after initial CABG. The CAAs had continued to grow gradually after the first CABG, becoming giant. Patients with CAAs require carefully tailored management.

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**Author contributions**

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**References**

1. Topaz O, Rutherford MS, Mackey-Bojack S, et al. Giant aneurysms of coronary arteries and saphenous vein grafts: angiographic findings and histopathological correlates. *Cardiovasc Pathol* 2005; 14(6): 298–302.
2. Esposito L, Di Maio M, Silverio A, et al. Treatment and outcome of patients with coronary artery ectasia: current evidence and novel opportunities for an old dilemma. *Front Cardiovasc Med* 2021; 8: 805727.
3. Li D, Wu Q, Sun L, et al. Surgical treatment of giant coronary artery aneurysm. *J Thorac Cardiovasc Surg* 2005; 130: 817–821.
4. Kawsara A, Nunez Gil IJ, Alqahtani F, et al. Management of coronary artery aneurysms. *JACC Cardiovasc Interv* 2018; 11: 1211–1223.
5. Syed M and Lesch M. Coronary artery aneurysm: a review. *Prog Cardiovasc Dis* 1997; 40: 77–84.
6. Pahlavan PS and Niroomand F. Coronary artery aneurysm: a review. *Clin Cardiol* 2006; 29: 439–443.
7. Diaz-Zamudio M, Bacilio-Perez U, Herrera-Zarza MC, et al. Coronary artery aneurysms and ectasia: role of coronary CT angiography. *Radiographics* 2009; 29(7): 1939–1954.
8. Markis JE, Joffe CD, Cohn PF, et al. Clinical significance of coronary arterial ectasia. *Am J Cardiol* 1976; 37(2): 217–222.
9. Yamamoto H, Ito Y, Isogai J, et al. Immunoglobulin G4-related multiple giant coronary artery aneurysms and a single left gastric artery aneurysm. *JACC Case Rep* 2020; 2(5): 769–774.
10. Ueyama K, Tomita S, Takehara A, et al. [A case of surgical treatment for cardiac tamponade caused by a ruptured coronary aneurysm accompanied by a coronary artery-pulmonary artery fistula]. *Kyobu Geka* 2001; 54(1): 70–75.