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Successful Surgical Treatment of Giant Coronary Artery Aneurysm and Concomitant Coronary Artery Fistula to the Pulmonary Artery

Keito Suzuki, MD,[1] Naoyuki Kimura, MD, PhD,[1,2] Akira Sezai, MD, PhD,[1] Satoshi Unosawa MD, PhD,[1] Makoto Taoka, MD, PhD,[1] Shunji Osaka, MD, PhD,[1] Masashi Tanaka, MD, PhD.[1,2]

[1]Department of Cardiovascular Surgery, Nihon University School of Medicine, Tokyo, Japan; [2]Department of Cardiovascular Surgery, Saitama Medical Center, Jichi Medical University, Saitama, Japan

ABSTRACT

Giant coronary artery aneurysm (GCAA) combined with coronary artery fistula to the pulmonary artery (PA) is rare. A 79-year-old man was accidentally discovered with GCAA. He was operated on by use of aneurysmorrhaphy, and closure of the fistulae was performed. Because ischemic changes appeared, coronary artery bypass grafting was done. The postoperative course was uneventful, and the patient was discharged on postoperative day 14. We report here a case of GCAA with a size of 66 × 52 mm in diameter associated with a fistula formation into the PA. It is one of the largest sizes of GCAA that has occurred after fistula formation.

INTRODUCTION

Coronary artery aneurysm (CAA) is occasionally detected during the coronary angiography (CAG) and is defined as a localized dilatation of a coronary artery segment more than 1.5-fold compared with adjacent normal segments [Abou Sherif 2017]. The incidence of CAA has been reported to range from 0.3% to 5.3% in patients who undergo CAG [Abou Sherif 2017]. Although no consensus definition exists, coronary artery aneurysms are considered "giant," if the diameter exceeds 20 mm, 40 mm, 50 mm, or quadruple the reference diameter [Crawley 20144]. The reported incidences of giant coronary artery aneurysm (GCAA) decrease, and the prevalence of GCAA >50 mm was reported to be 0.02% [Crawley 20144]. Coronary artery fistula is another rare anatomical abnormality in which coronary arterial flow is shunted into a cardiac chamber or great vessels, with a reported incidence ranging from 0.1% to 0.8% of adult population [Cao 2015]. GCAA combined with coronary artery fistula to the pulmonary artery (PA) is rare [Tokunaga 2010; Morita 2012; Chiang 2014; Cao 2015], and its clinical manifestations have not been fully understood. We report here a case of GCAA with a size of 66 × 52 mm in diameter associated with a fistula formation into the PA.

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Correspondence: Masashi Tanaka, MD, PhD, Department of Cardiovascular Surgery, Nihon University School of Medicine, 30-1 Oyaguchi-kamimachi Itabashi-ku Tokyo, 173-8610, Japan; +81-3-3972-8111; fax: +81-3-3955-9818 (e-mail: tanaka.masashi@nihon-u.ac.jp).

CASE REPORT

A 79-year-old man who presented with a protrusion of the right-sided superior mediastinum on a chest x-ray was referred to our hospital. A computed tomography coronary angiography showed 2 aneurysms adjacent to the left ventricle, a larger left-sided 71-mm mass and a smaller right-sided 10-mm mass (Figure 1A). CAAs, including GCCA, was suspected. The patient was hospitalized for further evaluation. The patient did not present with shortness of breath or dyspnea on exertion. His past medical history and family history were unremarkable with nothing suggestive of Kawasaki disease. Transesophageal echocardiography (TEE) confirmed a 66-×-52-mm mass adjacent to the left ventricle with a diastolic blood flow from the proximal portion of the left anterior descending artery (LAD). TEE failed to demonstrate direct communication between the large CAA and any cardiac cavities (Figure 1B). TEE also showed that there were multiple coronary artery fistulae shunted to the main PA (Figure 1C). The shunt rate was not measured. TEE showed normal left ventricular wall motion with ejection fraction of

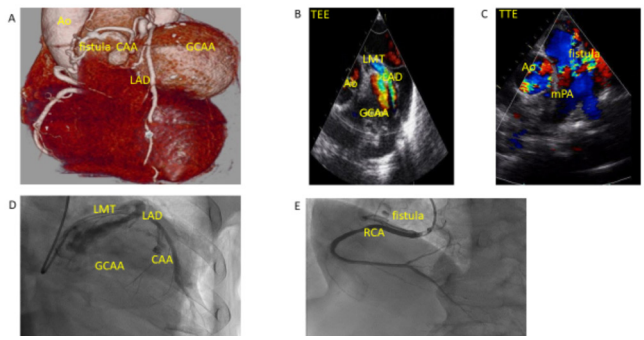

Figure 1. A, Computed tomography coronary angiography showed a large 71-mm aneurysm adjacent to the LAD and a concomitant smaller aneurysm. B, TEE showed a 66-×-52-mm aneurysm adjacent to the left ventricle with no direct communications to the cardiac cavities. Blood flow from the proximal portion of the LAD into the aneurysm was delineated during diastole. C, TEE showed multiple fistulae to the main PA. D, CAG showed blood flow from the proximal portion of the LAD into the aneurysm. E, CAG showed multiple fistulae entering from the RCA to the main PA.

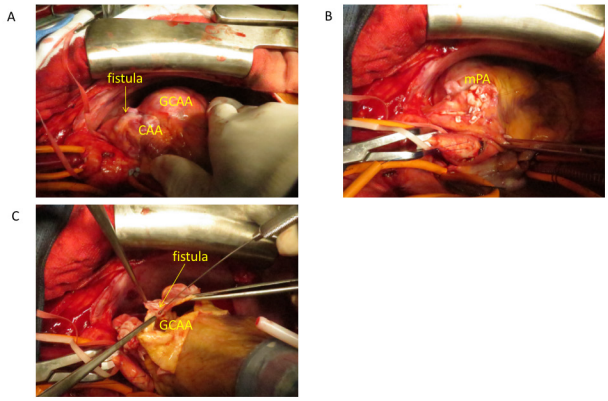

Figure 2. Intraoperative image. A, GCAA adjacent to the left ventricle and a smaller CAA. B, Opening the mPA revealed numerous fistulous vessels from coronary arteries. C, Fistula to the mPA was visualized after incision of the GCAA.

64%. CAG showed a large aneurysm adjacent to the LAD to which blood flow was supplied from the proximal LAD during diastole (Figure 1D). CAG also showed multiple fistulae entering from the right coronary artery (RCA) to the main PA (Figure 1E). GCAA and concomitant smaller CAA associated with coronary artery fistulae to the main PA were diagnosed. Surgical treatment was scheduled urgently for GCAA and concomitant pulmonary fistulae.

During induction of general anesthesia, hemodynamics status was stable. Median sternotomy was performed, and inspection showed a large pulsatile GCAA adjacent to the left ventricle and showed a concomitant smaller CAA with a dilated fistulous vessel adjacent to the main PA (Figure 2A). Cardiopulmonary bypass was established with ascending aorta cannulation, bicaval venous cannulations. The patient was cooled to a tympanic temperature of 32 C. The ascending aorta was clumped, and cardioplegic arrest was obtained via antegrade coronary perfusion, and then retrograde perfusion through the coronary sinus was used intermittently. First, the main PA was opened longitudinally, and multiple fistulae connecting to the main PA were identified (Figure 2B). All these fistulae were ligated, and the PA was closed with a 4-0 Prolene polypropylene running suture. The GCAA was subsequently incised, and the inflow path from the LAD and the outflow pass connecting to the main pulmonary artery (mPA), which had not been detected during preoperative imaging tests, were identified (Figure 2C) and ligated. After completion of aneurysmotomy of GCAA and ligation of multiple fistulae to the PA, the ascending aorta was declamped, and the heart resumed beating spontaneously. During weaning from cardiopulmonary bypass, TEE revealed a decreased left ventricular wall motion in the anteromedial septal region, and electrocardiogram showed ST segment depression. Although the possibility of air embolism was considered, we performed coronary artery bypass grafting (CABG) to the LAD by using a saphenous vein graft because of fear of ligation of the LAD. Urgent coronary revascularization improved left ventricular wall motion and electrocardiogram abnormality.

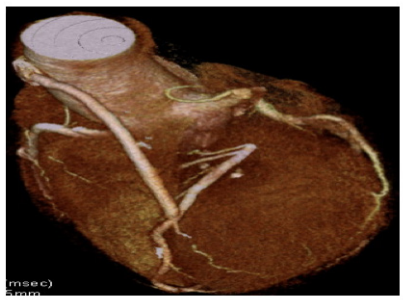

Figure 3. Postoperative computed tomography coronary angiography showed that GCAA and smaller CAAs disappeared and that the saphenous vein graft bypassed to the LAD flow was patent.

Pathological examination of the resected GCAA specimen showed atherosclerosis of aneurysmal wall, including accumulation of foam cells, cholesterin crystals, and calcification, without the normal 3 layers of an arterial wall. Histologically, GCAA was a pseudoaneurysm and was considered to develop at the site of coronary arterial fistula. A postoperative computed tomography coronary angiography showed deletion of CAAs and patent saphenous vein graft (Figure 3). The postoperative course was uneventful, and the patient was discharged on postoperative day 14.

DISCUSSION

CAA is a rare coronary abnormality, and a few previous literatures reported the GCAA associated with coronary artery fistula to the PA [Tokunaga 2010; Morita 2012; Chiang 2014; Cao 2015]. Hirose and colleagues reported that the incidence of CAA associated with fistula was found in only 0.02% of patients who had undergone coronary angiography [Hirose 1999]. Li and colleagues also reported that CAA was found in 5.9% of patients with congenital coronary artery fistula, which was found in only 0.2% of patients who had undergone heart surgery [Li 2005]. CAA can be congenital or acquired, etiologically. The most frequent cause of acquired aneurysms is atherosclerosis, although other causes include Takayasu arteritis, Kawasaki disease, IgG4-related coronary periarteritis, and injury during percutaneous coronary intervention [Barr 2016]. The RCA is most often affected (89%), whereas the proximal LADs are exceedingly rare, although one 7-cm giant CAA from a septal branch was reported [Crawley 2014]. To the best of our knowledge, the size of the largest GCAA was 118 × 136 mm in diameter, which was reported by Zhaoping et al [Zhaoping 2015]. A large-sized CAA in excess of 70 mm in diameter shown in this case was rare, and there has been limited information concerning clinical presentation and treatment of large-sized GCAAs. The present case did not have any evidence of collagen vascular disease or Kawasaki disease. We consider that the CAAs, including GCAA, developed in association with atherosclerosis on the site of congenital fistula formation.

Most of GCAAs are asymptomatic; however, patients with GCAA can present with angina pectoris, sudden death, fistula

formation, pericardial tamponade, compression of surrounding structures, or congestive heart failure [Crawley 2014]. Therefore, a variety of imaging techniques have been used to identify CAAs. Noninvasive methods, such as echocardiography, computed tomography, and magnetic resonance imaging, can detect some of these aneurysms. Coronary angiography remains the most important imaging modality, because it provides information about the size, morphology, location, number of lesions, and communication to the cardiac chambers or vessels. In our patient, the inflow path from the proximal LAD to the GCAA was detected by echocardiography and CAG; the output path to the mPA was not detected by imaging modalities preoperatively.

Considering the risk of rupture, or subsequent myocardial ischemia and thrombotic events, GCAA is usually managed surgically [Hirose 1999; Li 2005; Chiang 2014; Barr 2016]. Surgical treatments include aneurysmotomy with [Li 2005; Chiang 2014; Barr 2016] or without [Li 2005; Morita 2012; Cao 2015] coronary revascularization. Coronary surgical revascularization is performed on the basis of preoperative evaluation for concomitant coronary ischemia. Our case did not have stenosis lesions in the downstream LAD; however, coronary ischemia in the LAD area was suspected intraoperatively after surgical correction of the GCAA. Adding coronary artery bypass grafting successfully improved coronary ischemia, suggesting that air thrombosis or ligation of LAD might have occurred during aneurysmotomy. In cases in which GCAA is accompanied by coronary artery fistula to the PA, all fistulous connections should be ligated [Tokunaga 2010; Morita 2012; Chiang 2014; Cao 2015]. Opening the main PA would help identify fistulous communication between the PA and aneurysm.

We experienced a very rare case of GCAA associated with a smaller CAA and coronary arterial fistula to the PA. The patient successfully underwent open surgical repair and concomitant coronary artery bypass grafting. Etiologically, GCAA was considered to develop at the site of coronary arterial fistula as a result of atherosclerotic change.

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