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CLINICAL REPORT

Giant coronary artery aneurysm with coronary arteriovenous fistula draining into the coronary sinus

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Abstract A 77-year-old patient suffering from a giant right coronary artery aneurysm with coronary arteriovenous fistula was admitted to our hospital. The fistula could not be documented preoperatively by computed tomography or coronary angiography but was documented intraoperatively by transesophageal echocardiography (TEE). However, TEE was unable to visualize the draining site of the fistula. Direct palpation by the surgeon ultimately confirmed that the fistula was draining into the coronary sinus. The fistula was closed and the volume of the aneurysm reduced by partial resection. The postoperative course of the patient was uneventful. Giant aneurysms occasionally displace cardiac structures. In such cases, combined imaging technologies, including TEE, may be needed for precise assessment of the giant aneurysm and fistula.

Keywords Coronary artery aneurysm · Coronary arteriovenous fistula · Transesophageal echocardiography

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Introduction

Coronary artery aneurysm (CAA) is a rare disease that occasionally occurs with coronary arteriovenous fistula (CAF). Giant aneurysms occasionally compress and displace cardiac structures, making precise assessment of the aneurysm for surgery difficult. We present a case of CAA accompanying CAF detected by transesophageal echocardiography (TEE) during surgery.

Case report

A 77-year-old hypertensive woman with no history of Kawasaki's disease or chest trauma was admitted to a small private hospital due to nausea. Her blood pressure (BP) was 88/64 mmHg and heart rate (HR) was 135 beats per minute (bpm). An electrocardiogram showed paroxysmal supraventricular tachycardia. Bepridil hydrochloride administration was initiated, and the patient was hospitalized. Although her hemodynamic condition improved to demonstrate normal sinus rhythm with a HR of 90 bpm and BP of 130/60 mmHg, her chest roentgenogram showed an abnormal shadow adjacent to the left ventricle with a positive silhouette sign. Transthoracic echocardiography revealed a 72 × 68-mm^2 cystic structure adjacent to the left chamber of the heart. Contrast-enhanced computed tomography (CT) documented a CAA that was 80 × 70 × 80 mm^3 (Fig. 1). The patient was referred to our hospital for surgical treatment.

Coronary angiography (CAG) confirmed a giant aneurysm of the peripheral segment of the right coronary artery (Fig. 2). The right coronary artery was dilated to more than twice the width of the left coronary artery. Neither CT nor CAG detected any draining vessels, which indicated CAF.

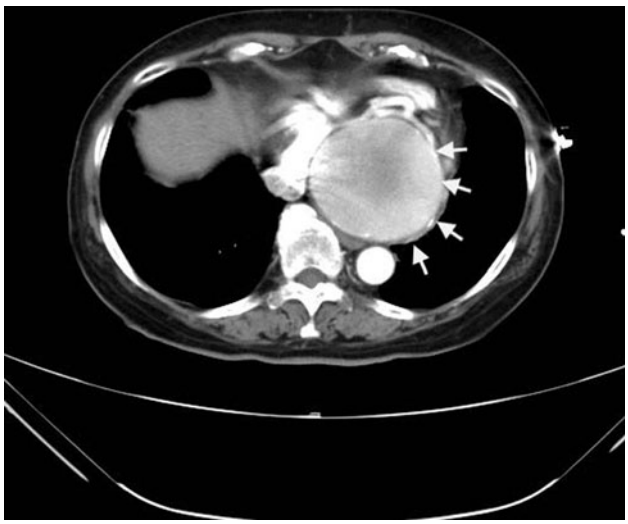

Fig. 1 Computed tomography (CT) image. The image shows a giant mass (*white arrows*) enhanced by contrast in front of the descending aorta. The wall of the aneurysm contains calcification spots. A tortuous vessel courses ventrally to the aneurysm

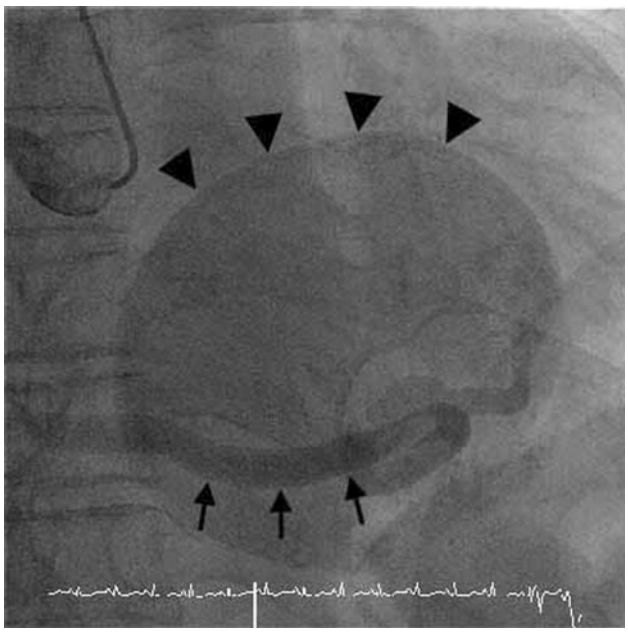

Fig. 2 Coronary angiogram. The image shows the dilated right coronary artery (*arrows*) and giant aneurysm (*arrowheads*). Draining vessels are not visualized

Resection of the giant aneurysm under cardiopulmonary bypass (CPB) via median sternotomy was scheduled.

Following the induction of general anesthesia using ketamine, propofol, remifentanil, and rocuronium, the trachea was intubated with an endotracheal tube (internal diameter 7.0 mm). A TEE probe was inserted through the esophagus, and a pulmonary artery catheter was inserted via the right internal jugular vein. The cardiac index following anesthesia induction was 2.0 L min^{-1} m^{-2}. There was an increase in oxygen saturation from 72% in the superior vena cava to 81% in the pulmonary artery, which indicated the existence of a shunt, such as an atrial septal defect (ASD). TEE examination confirmed the absence of ASD, ventricular septal defect, and patent ductus arteriosus. It also revealed a 68 × 52-mm^2 giant aneurysm with a thick wall possessing an irregular inner surface. Color flow Doppler examination showed low-velocity, swirling blood flow inside the aneurysm. It also documented a pulsatile inward blood signal and an outward blood signal 8 mm to the left of the inward signal (Fig. 3). Although displacement of the heart structure by the aneurysm made it difficult to determine where the outward blood flow was draining, a giant CAA with CAF was suspected.

The aneurysm was incised following CPB establishment and cardiac arrest, and an ostium from the posterior descending artery and an outlet ostium connecting to the coronary sinus were detected with a probe by the surgeon, revealing the CAF draining into the coronary sinus. After both ostia were ligated with pledgeted 2-0 Nespolen (Asflex, Tokyo, Japan) sutures, the volume of the aneurysm was reduced by partial resection of the wall and the aneurysm closed. The patient was separated from CPB without incident. The post-bypass TEE examination revealed no wall motion abnormality of the left ventricle, but did not delineate the interior of the aneurysm because of the interposition of air resulting from the opening of the aneurysm. Postoperative contrast-enhanced CT showed no enhancement of the aneurysm. The postoperative course of the patient was steady, and she was discharged on postoperative day 20. The histopathology of the excised aneurysm wall displayed widespread fibrosis with hyalinization, corresponding with the finding of an atherosclerotic change of the coronary artery.

Discussion

Coronary artery aneurysm is an uncommon disease, with an incidence rate of between 1.5 and 5% [1]. The main cause of CAA is atherosclerotic change in the coronary arteries [1], but it is also caused by other conditions, such as Kawasaki's disease, Takayasu's disease, infectious endocarditis, or iatrogenic events, including stent implantation [1]. According to a review of 22 reports, a giant aneurysm is usually defined as a coronary artery with a diameter of >2 cm [2].

CAF is also a rare heart anomaly that occurs in 0.1–0.2% of patients undergoing CAG [3, 4]. It is usually congenital, but on rare occasions is acquired after chest trauma or medical episodes, such as endomyocardial

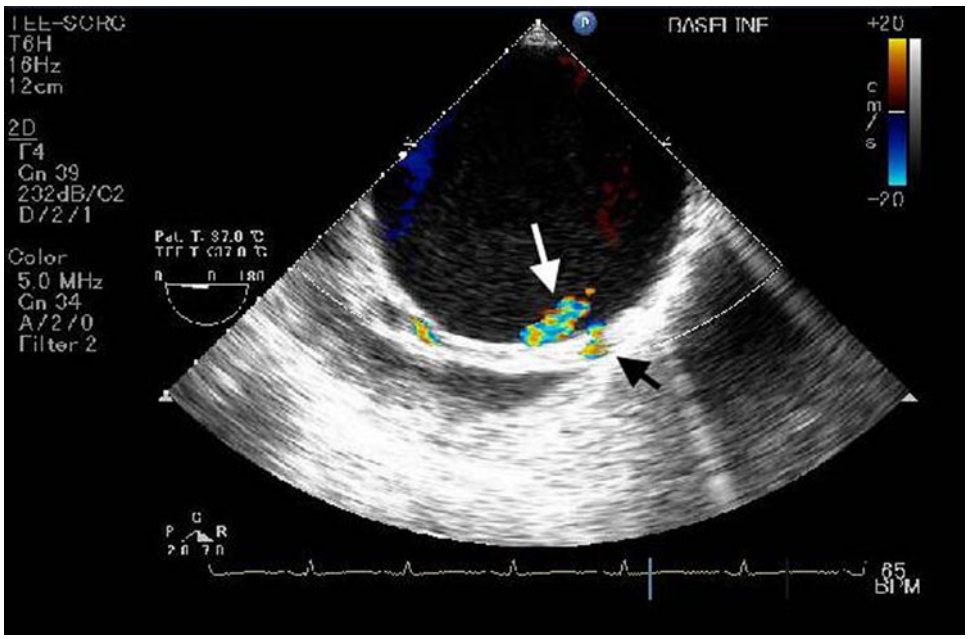

Fig. 3 Image of the aneurysm obtained by intraoperative transesophageal echocardiography. The color flow Doppler interrogation shows the inward (*white arrow*) and outward (*black arrow*) blood flow

biopsy [5]. Congenital CAF arises from incomplete obliteration of the primitive myocardial sinusoids during embryonic development [5, 6]. The feeding vessels originate from the right coronary artery in 50% of patients, the left coronary artery in 42%, and from both the left and right coronary artery in 5% of patients [4]. The usual drainage sites are the right atrium in 26% of patients, the pulmonary artery in 17%, and the coronary sinus in 7% of patients [4].

CAA also occurs in approximately 15–19% of patients with CAF [1, 3]. The reason for this is that a fistula consists of a nidus of vessels containing fragile smooth muscle, which is susceptible to constant exposure to arterial pressure and much blood flow, resulting in dilation and aneurysmal change with age [7]. Makaryu et al. [7] reported a case of giant CAA with CAF draining into the coronary sinus. They concluded that the fistula was formed by direct erosion and rupture of the aneurysm into the coronary sinus due to progressive atherosclerosis, with evidence of calcified walls of the aneurysm and irregularly shaped ostia of the inlet and outlet vessels [6]. With regard to our case, the patient had no history of chest trauma, cardiac surgery, or coronary intervention. The shapes of both ostia were smooth, and the aneurysm was not connected directly to the coronary sinus. Hence, we concluded that she had congenital CAF and developed a giant CAA. The atherosclerotic change of the CAA wall may have resulted from long-term exposure to arterial pressure.

Although there are some non-invasive tools for the diagnosis of CAA, such as echocardiography, CT, and magnetic resonance imaging, CAG remains the gold standard that provides information on the size, shape, and location of a CAA [7]. CAG has also been considered to be the gold standard for the identification of fistula origin in terms of CAF diagnosis. However, some reports show that CAG is limited in its ability to determine the course and drainage site of fistulas due to the overlap of adjacent structures; they also suggest that TEE provides additional information on the drainage site that CAG cannot obtain and that TEE could be complementary to angiography in the assessment of CAF [5, 8, 9].

Based on CT and CAG findings, we initially did not diagnose the patient with a fistula. Intraoperative TEE revealed the outlet head that CAG did not detect; however, we were unable to confirm the structure to which it was connected. This is clearly due to the inappropriate dimensional relationship between the TEE transducer in the esophagus and the cardiac structures that were compressed and displaced by the giant aneurysm. In our case, the coronary sinus was the exit site where the fistula was draining and we were unable to visualize it because of the anatomical feature mentioned above. If the coronary sinus had been delineated, we may have been able to see that it was dilated due to the blood flow through the fistula, suggesting the patient had persistent left superior vena cava; this would have been a clue for considering the presence of CAF. Although TEE is complementary to techniques such as CAG, a multimodal approach, including CT, angiography, TEE, and even direct palpation by surgeons, may be needed for precise assessment of the fistula and aneurysm when the CAA is very large, as it was in our case.

Complications of CAF include thrombosis, embolism, congestive heart failure, atrial fibrillation and other arrhythmias, endocarditis, and the rupture of the fistula [8]. Because the natural course of CAA with CAF is unclear, the management of patients is controversial [5, 7]. Rupture

occurs quite rarely but does happen on occasion [7]. Most studies have found that symptoms and complications increase with age, and surgical intervention before the development of symptoms is recommended [5]. Surgical correction of an isolated CAF is associated with low surgical mortality [5].

We treated this patient using surgery because she had symptoms of paroxysmal supraventricular tachycardia. Transcatheter nonsurgical embolization using coils and balloons for CAF has been reported [8], but we did not utilize this technique because we were not conversant with it. It may be preferred for patients facing high surgical risks; however, unsuccessful attempts in patients with giant aneurysms have been reported [6]. We agree with Inoue et al.'s suggestion in which surgical correction under CPB with cardiac arrest is recommended, which makes it possible to inspect the afferent and efferent vessels clearly from the inside [7].

In conclusion, we report a case of a patient with a giant CAA with CAF draining into the coronary sinus who was treated with surgical correction under CPB. An aneurysm that is very large could compress the cardiac structures and change the anatomical dimensions, which occasionally makes it difficult to detect the draining site of the fistula. Multimodal, combined imaging technologies, including TEE, should be used for precise assessment of a giant aneurysm.

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