Rupture of an Aneurysm Resulting From a Coronary Artery Fistula

— A Case Report —

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A rare case of coronary-to-pulmonary artery fistulas associated with formation of a saccular aneurysm that ruptured into the pericardium occurred in a 69-year-old female who had experienced an episode of unconsciousness 3 months earlier and who suffered a second episode. She was diagnosed as having a cardiac tamponade caused by rupture of a coronary artery aneurysm formed by a left coronary artery–pulmonary artery fistula. The hemorrhage stopped after pericardial drainage. She was referred for surgical treatment of the aneurysm and suture closure of the afferent coronary artery into the aneurysm, transpulmonary closure of the fistulas, and aneurysmorrhaphy. There was adhesion between the aneurysm and pericardium. Her postoperative course was uneventful and she has remained well for 4 months after the operation. (Circ J 2003; 67: 551–553)

Key Words: Coronary artery aneurysm; Coronary artery fistula; Rupture

Coronary artery fistula (CAF) and coronary artery aneurysm (CAA) are being recognized more often with the improvement in diagnostic techniques, such as selective coronary arteriography and echocardiography. Krause first described CAF in 1865 as characterized by the involved coronary artery having a normal origin from the aorta with a fistulous communication with the atria or ventricles, or with the pulmonary artery (PA). Although many CAF with CAA have been reported, it is quite rare that patient survives a rupture and, additionally, the progress of the present case is also unusual.

Case Report

A 69-year-old female with a history of unconsciousness 3 months earlier was referred for management of the syncope. On admission, she was conscious, her heart rate was 98 beats/min and blood pressure was 80/64 mmHg. There was no continuous murmur audible. Chest radiography showed an enlarged cardiac silhouette and a bulge of the left cardiac border next to the left PA (Fig 1). An electrocardiogram showed normal sinus rhythm, no significant ST-T changes, and low voltage in all the leads. An echocardiogram revealed massive pericardial effusion and a mass, with a maximum diameter of 4 cm, located at the left side of the PA. Computed tomography (CT) scan showed an enhanced mass with a maximum diameter of 3 cm originating from the left coronary artery (LCA) (Fig 2). Pericardial drainage was performed under local anesthesia for cardiac tamponade and 470 ml of blood was withdrawn. Thereafter, hemorrhage from the pericardial drainage stopped and selective coronary arteriography (CAG) (Fig 3) was immediately performed, which demonstrated a meandering CAF.
with a large saccular aneurysm without extravasation originating from the left anterior descending (LAD). Minor fistulas from the right coronary artery were also detected and all the fistulas from both coronary arteries drained into the main PA.

The patient was diagnosed as having a ruptured CAA with multiple CAF, but surgery was not performed immediately after CAG because the pericardial bleeding had ceased and melena originating from the colon had occurred. Therefore, an elective operation through a median sternotomy was planned for 1 month hence.

A 3×4 cm CAA was located on the left side of the PA (Fig 4) with adhesion between the aneurysm and the pericardium, but no rupture site. A slight thrill could be felt in the CAA and the PA. Under standard cardiopulmonary bypass the PA was opened and perfused with antegrade cold blood cardioplegia, which exposed the openings of the fistulae. A total of 4 orifices were closed from inside the PA using 5-0 prolene. Bleeding from the PA wall then ceased completely, despite the cardioplegic perfusion. Following closure of the PA wall, the aneurysm was opened and was found to be full of both old and fresh thrombi, which were removed, and the 2 orifices of the filling arteries were closed using 5-0 prolene continuous suture. Additional cardioplegic solution was perfused through the aortic root, but there were no further openings to the aneurysm. Redundant aneurysm wall was excised and aneurysmorrhaphy was performed. After evacuation of residual air, cardiopulmonary bypass was discontinued without any problems.

Postoperative recovery was uneventful with respect to hemodynamic status. CT scan showed an non-enhanced area of low attenuation in the left side of the PA and CAG showed that both CAA and CAF had disappeared (Fig 5). Pathological examination of the aneurysm wall showed atherosclerotic changes in the intima and media, and degenerative and inflammatory changes in the adventitia. The patient remains well at 4 months after the operation.

Discussion

CAA is a relatively rare disease that may cause angina, myocardial infarction, or sudden unexpected death from thrombosis, embolization or rupture. CAF is also an uncommon congenital heart defect (0.2% of congenital cardiac anomalies and 0.2–1% of all selective coronary arteriography cases), but is easily treated with surgery. The fistula usually originate from the RCA (55%), but may arise from the LCA (35%), both coronary arteries (5%), or an anomalous single coronary artery (3%). The low-pressure chambers are the usual drainage sites: the right ventricle, 42.5%; the right atrium, 34%; the pulmonary artery, 15%; the left atrium, 5%; the left ventricle, 3.5%. There have been many reports of CAA concomitant with CAF, but those with rupture and cardiac tamponade are very rare. We speculate that in the present case, congenital CAF were already extant and the CAA was secondary, probably as a result of the atherosclerotic changes of the walls of the CAF, because the patient did not have a history of chest...
trauma or have inflammatory symptoms similar to aortitis. It was rupture of the atherosclerotic aneurysm that resulted in cardiac tamponade.

Early treatment, especially in the asymptomatic young patient, should prevent the appearance of later symptoms or complications. Although Konno et al suggested the surgical indications in 1973,17 the surgical risk under current open-heart techniques appears to be considerably less than the potential development of serious complications.18 Fortunately, the rupture site of the aneurysm was closed by adhesion to the pericardium in this patient, but if the bleeding from the aneurysm had not stopped, her prognosis would have been poor. Therefore, surgical treatment of CAF should be undertaken when the diagnosis is made, either without the use of cardiopulmonary bypass or, in the view of the current improved techniques, employed routinely in most instances. We performed transpulmonary closure of the coronary artery–PA fistula, suture closure of the afferent coronary artery to the aneurysm, and aneurysmorrhaphy. Because the distance between the LAD and the aneurysm was short, aortocoronary bypass grafting was not required. Perfusion of cardioplegia during the cross-clamping period is extremely important because it exposes the orifices of the fill-in arteries to the aneurysm and the draining vessels to the PA. However, if an antegrade cardioplegia does not provide adequate myocardial protection, retrograde cardioplegia should be used.

References