

Letter to the Editor

A case of fistulous tracts from all coronary arteries to pulmonary trunk, right ventricle, left atrium, bronchial arteries and aorta, co-existing fistulas between bronchial arteries and pulmonary arteries



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Coronary artery fistula (CAF) is an anomalous connection between a coronary artery and another vessel or cardiac chamber. Most CAFs commonly involve a single communication (93%) or two communications (6%) [1]. CAF can steal downstream blood flow from the coronary artery, resulting in hypo-perfusion and cardinal symptoms [2]. We report a patient with fistulous tracts from all coronary arteries to pulmonary trunk, right ventricle, left atrium, bronchial arteries and aorta, co-existing fistulas between bronchial arteries and pulmonary arteries without significant coronary artery disease. The patient did not complain chest pain after medical supportive care, and Tc99m-MIBI-SPECT revealed no evidence of ischemia from CAFs.

A 65-year-old woman was referred to the emergency department because of new onset resting chest pain. She was diagnosed with bronchiectasis due to hemoptysis 10 years earlier. Her vital signs were normal except for tachypnea (respiratory rate 24/minute). Grade III systolic ejection murmur which radiated to neck was auscultated at aortic area. Electrocardiogram showed normal sinus rhythm. The admission chest radiograph showed bronchiectasis in both lower lung fields without interval change. To further evaluate the resting pain and systolic murmur, transthoracic echocardiography was performed, and it showed normal heart function without aortic valve disease. We decided to perform coronary angiography for recurrent squeezing chest pain on substernal area. Coronary angiography (Fig. 1) showed fistulous arteries from all

coronary arteries to pulmonary trunk without significant coronary artery disease. Left main (LM) coronary artery had mild arteriovenous (AV) fistula to pulmonary trunk. Left anterior descending (LAD), left circumflex (LCX), and right coronary artery (RCA) had big AV fistula to pulmonary trunk. Three-dimensional (3D) contrast-enhanced coronary CT angiography (Fig. 2) showed pulmonary artery fistulas (CPAFs) between RCA and LCA, and pulmonary artery (PA). Stream of contrast media from small communication (diameter < 0.5 cm) between coronary artery and anterior portion of right ventricular outflow tract (RVOT) were noted and communication with left atrium was also noted. Remarkable dilatation in the proximal RCA and left coronary artery (LCA) with development of collateral blood supply and small thrombi within the dilated LCX branch was observed. There were bronchiectasis and bronchial artery aneurysms in the right middle lobe and left lingual segment. Aorta had fistulous tracts with LAD coronary artery, and bronchial arteries also had fistulous tracts with pulmonary arteries. Therefore, she was identified with congenital multiple CAFs as none of acquired complications was suspected. To evaluate ischemic insult from CAFs, technetium 99 m sestamibi single emission computed tomography (Tc99m-MIBI-SPECT) (Fig. 3) was conducted, but no significant perfusion defect was observed even after adenosine stress test. After supportive care with vasodilator (trimetazidine) and antiplatelet (triflusal) therapy, the patient has been asymptomatic until now.

The incidence of CAF in overall population is presumed to be about 0.002%, and the incidence in angiography is estimated to be about 0.05–0.12% [3]. The pathogenesis of CAFs remains uncertain. One study showed that CAFs were communications of immature vessels surrounding the pulmonary artery and ductus arteriosus with left sixth arch artery [4]. In the embryonic development of the coronary artery, the epicardial coronary plexus formed a ring around the arterial trunk and then developed into many major branches. Although the other supernumerary vessels regress spontaneously, persistence remnant of these vessels might be the coronary–pulmonary artery fistulas [5,6].

Most fistulas have a single communication and multiple fistulas are rare condition [7], reported incidence of multiple fistulas is 2% in the patients who have CAFs [8]. This case is the first case reporting extremely rare pattern of multiple fistulas between all major coronary arteries, right ventricle, left atrium and bronchial arteries. The treatment of CAFs is needed when CAFs cause myocardial ischemia, left ventricular dysfunction, congestive cardiac failure and symptomatic arrhythmia. In this case, surgical ligation or catheter closure [9] was not yet

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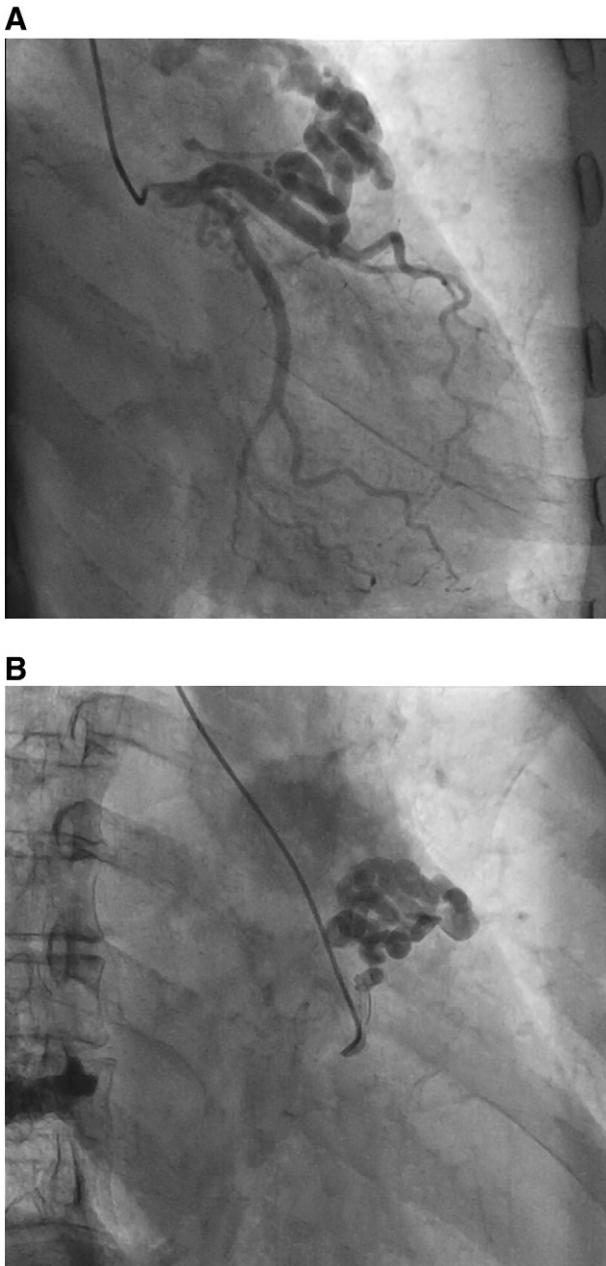


Fig. 1. Coronary angiography showed fistulous arteries from all coronary arteries to pulmonary trunk. (A) Coronary artery fistulas between left main, left anterior descending, left circumflex coronary arteries and pulmonary trunk. (B) Coronary artery fistula between right coronary artery and pulmonary trunk.

indicated, as Tc99m-MIBI-SPECT demonstrated no significant perfusion defect even after adenosine stress test. Multiple fistulas and small thrombi within the dilated LCX branch also supported the medical therapy as sometimes the thrombi could induce spontaneous closure of CAFs. We prescribed an anti-platelet agent, triflusal, to prevent ischemic events by the thrombi. If her symptoms suggesting congestive heart failure, arrhythmia, sudden death, infective endocarditis and myocardial infarction occur [10], we should consider surgical ligations rather than catheter closure because of high fistula flow, multiple communications, and multiple terminations.

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