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Anomalous origin of the left coronary artery from the pulmonary artery: A rare case in an adult

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A 46-year-old man presented at the outpatient cardiology clinic with a 5-year history of paroxysmal atrial fibrillation. During this period, he suffered several palpitations triggered by alcohol or fatigue, with cardioversion of atrial fibrillation to sinus rhythm through intravenous administration of amiodarone. This time he presented with palpitation for 3 days, without chest pain, shortness of breath, cyanosis, or edema. The electrocardiogram revealed atrial fibrillation. The transthoracic echocardiogram revealed left atrial enlargement (48 mm), near upper limit of left ventricle (54 mm), right atrial enlargement (41 mm), left ventricular ejection fraction of 57%, as well as mild to moderate mitral regurgitation, mild aortic regurgitation, and mild

tricuspid regurgitation. Unusually, the transthoracic echocardiogram revealed a dilated right coronary artery with an internal diameter of approximately 8 mm, originating from the ascending aorta (Figure 1A). While an abnormal blood flow directed towards the pulmonary artery (Figure 1B) could indicative of an anomalous connection between the pulmonary artery and the left coronary artery. Abnormal vascular structure with reversed flow detectable inside the interventricular septum on color doppler was observed (Figure 1C), suggesting the reversed flow from the right coronary artery to the left coronary artery through coronary collateral vessels. According to these image characteristics, the primary diagnosis of anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) was proposed. Furthermore, the patient confirmed the diagnosis through coronary computed tomography angiography (Figure 1D–E) and coronary angiography (Figure 1F), which both revealed left coronary artery anomaly originating from the pulmonary artery. The patient then underwent surgery by ligation of the abnormal opening of the left coronary artery and coronary artery bypass grafting to establish circulation from the aorta to the left anterior descending branch. Normal myocardial perfusion was restored after surgery. One month after surgery, the patient was asymptomatic. Physical examination showed no signs of heart failure, and the electrocardiogram revealed atrial fibrillation. The transthoracic echocardiogram revealed a residual 2 mm shunt bundle at the opening from the left coronary artery to the pulmonary artery. ALCAPA, also known as Bland–White–Garland syndrome, is considered a rare congenital anomaly of the coronary arteries, which occurs in 1 out of 300 000 live births. Without treatment, 90% of infants will die within the first year of life. Survival to adulthood for patients with ALCAPA syndrome is extremely rare [1]. We reported the case of a patient who survived 46 years without obviously left ventricular dysfunction. It is attributed to the collateral circulation between the left and right coronary arteries which ensured adequate blood supply to the heart. Despite this, chronic left ventricular subendocardial ischemia may occur, posing a high risk for sudden cardiac death due to malignant ventricular arrhythmias. Transthoracic echocardiogram provides great value as the first-choice method for the correct diagnosis of ALCAPA. Characteristic findings, such as a dilated right coronary artery, abnormal blood flow towards the pulmonary artery, and abnormal

vascular structure with reversed flow inside the interventricular septum, are key diagnostic indicators of ALCAPA. Early diagnosis and prompt surgical intervention are crucial for improving outcomes and preventing complications such as left ventricular dysfunction and sudden cardiac death [2].

Article information

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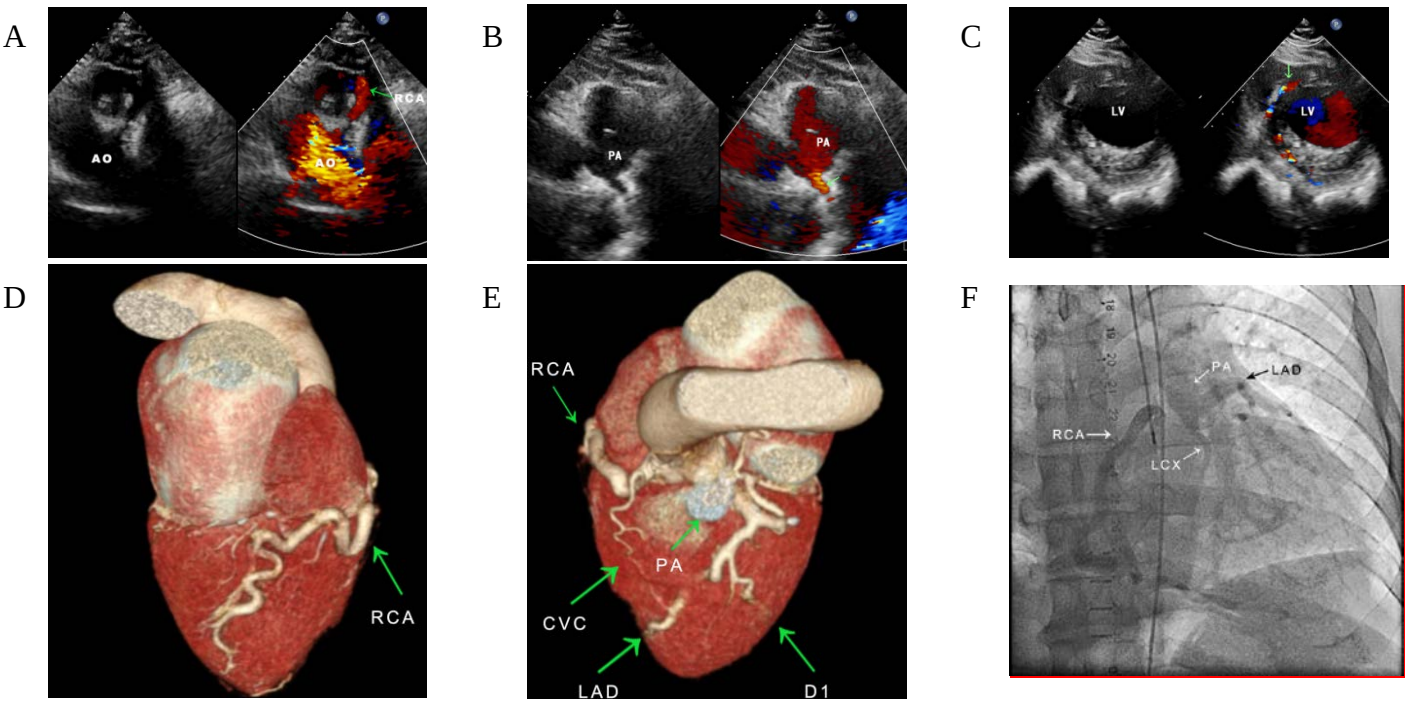

Figure 1. A. Transthoracic echocardiography revealed a dilated RCA originated from Ao. **B.** Color Doppler revealed an abnormal blood flow directed towards the PA. **C.** Color Doppler revealed turbulent flow inside the interventricular septum. **D.** Volume-rendered 3D coronary computed tomography revealed a dilated RCA. **E.** The LCX originating from the pulmonary trunk and the collateral circulation between the left and RCA. **F.** The baseline coronary angiography revealed that the thick RCA supplied blood to the tortuous left coronary artery *via* collateral circulation, with the left coronary artery opening into the pulmonary sinus at the root of the pulmonary artery

Abbreviations: Ao, ascending aorta; CVC, collateral vascular circulation; D1, diagonal branches 1; LAD, left anterior descending artery; LCX, left circumflex artery; LV, left ventricle; PA, pulmonary artery; RCA, right coronary artery