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

Lin Wang[1]*, Dong Cai[2]*, Shuya Zhao[2], Deren Kong[2], Xin Du[2], Miaomiao Jiao[1], Shanshan Lin[1], Mingwei Li[1], Jingyuan Mao[1]

[1]Department of Cardiology, 1st Teaching Hospital of Tianjin University of Traditional Chinese Medicine, National Clinical Research Center for Chinese Medicine Acupuncture and Moxibustion, Tianjin, China

[2]Internet Hospital of Tianjin Medical University General Hospital, Tianjin, China

*Both authors equally contributed to the study.

Correspondence to:
Jingyuan Mao, MD, PhD,
Department of Cardiology,
1st Teaching Hospital of Tianjin University of Traditional Chinese Medicine
No. 88, Changling Road, Tianjin, China,
phone: 022 2798 63 14,
e-mail: jymao@126.com

DOI: 10.33963/v.phj.100916

Received:
May 8, 2024

Accepted:
May 29, 2024

Early publication date:
June 3, 2024

A 46-year-old man presented at the outpatient cardiology center 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 palpitations lasting for 3 days, without chest pain, shortness of breath, cyanosis, or edema. An electrocardiogram revealed atrial fibrillation. A transthoracic echocardiogram showed left atrial enlargement (48 mm), near the upper limit of the 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). An abnormal blood flow directed towards the pulmonary artery (Figure 1B) could indicate an anomalous connection between the pulmonary artery and the left coronary artery. An abnormal vascular structure with reversed flow detectable inside the interventricular septum on color Doppler was observed (Figure 1C), which suggested the reversed flow from the right coronary artery to the left coronary artery through coronary collateral vessels.

Based on these image characteristics, the primary diagnosis of anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) was proposed. Furthermore, this diagnosis was confirmed by coronary computed tomography angiography (Figure 1D–E) and coronary angiography (Figure 1F), which both revealed a 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. An electrocardiogram revealed atrial fibrillation. The transthoracic echocardiogram showed 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 coronary arteries, which occurs in 1 out of 300 000 live births. Without treatment, 90% of infants 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 obvious left ventricular dysfunction. This is attributed to the collateral circulation between the left and right coronary arteries which ensures adequate blood supply to the heart. Despite this, chronic left ventricular subendocardial ischemia may occur, posing a high risk of sudden cardiac death due to malignant ventricular arrhythmias. Transthoracic echocardiogram provides great value as the first-choice method for the correct diagnosis

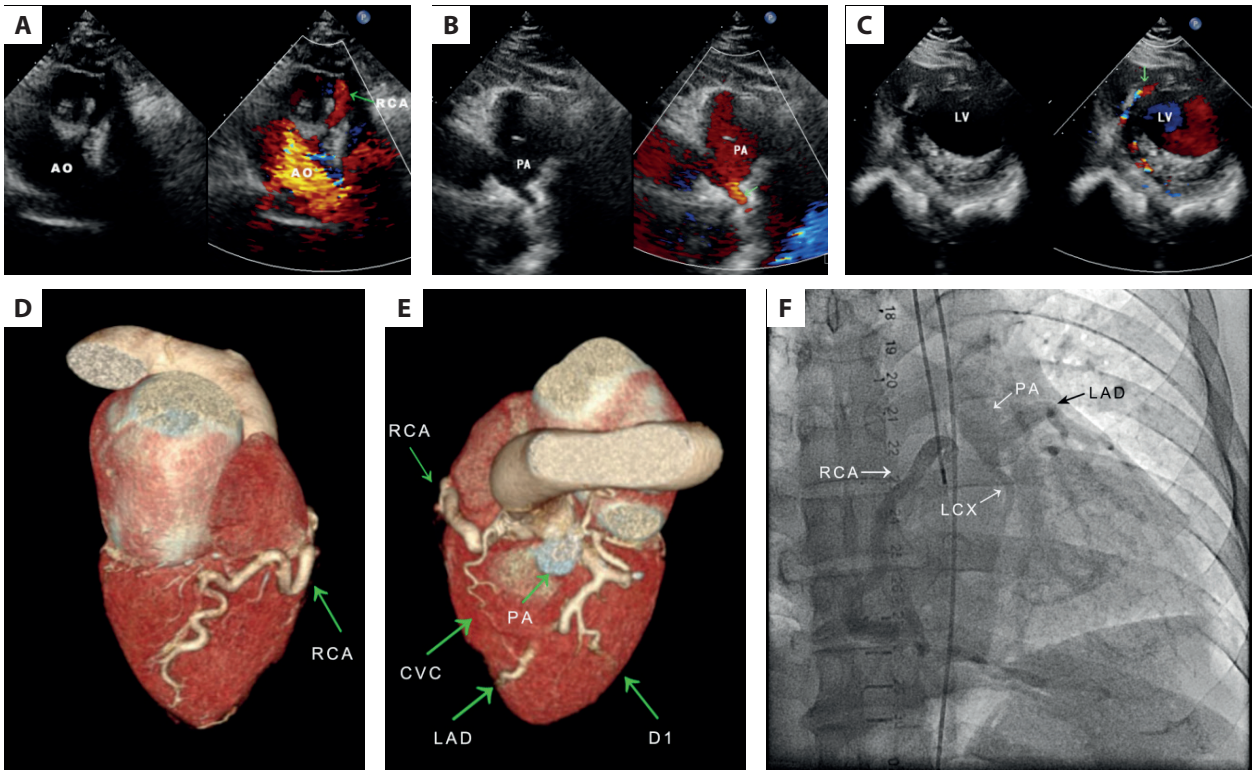

Figure 1. A. Transthoracic echocardiography revealed a dilated RCA originated from the Ao. **B.** Color Doppler showed abnormal blood flow directed towards the PA. **C.** Color Doppler revealed turbulent flow inside the interventricular septum. **D.** Volume-rendered 3D coronary computed tomography indicated a dilated RCA. **E.** The LCX originating from the pulmonary trunk and the collateral circulation between the left and right coronary arteries. **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

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

Conflict of interest: None declared.

Funding: This work was funded by the Innovation Team and Talents Cultivation Program of the National Administration of Traditional Chinese Medicine (No. ZYYCXTD-C-202203).

REFERENCES

1. Maron BJ, Doerer JJ, Haas TS, et al. Sudden deaths in young competitive athletes: analysis of 1866 deaths in the United States, 1980–2006. Circulation. 2009; 119(8): 1085–1092, doi: 10.1161/CIRCULATIONAHA.108.804617, indexed in Pubmed: 19221222.
2. Yalta K. Presence of an anomalous coronary artery: a potential risk factor for device-related late coronary ischemic syndromes following transcatheter aortic valve implantation? Kardiol Pol. 2023; 81(3): 322–323, doi: 10.33963/KP.a2022.0253, indexed in Pubmed: 36987940.