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A Malignant Course of Abnormal Origin of Right Coronary Artery and Left Circumflex Artery Arising from Left Coronary Sinus Presented with Chest Pain

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Abstract

Abnormal origin of the right coronary artery (RCA) from the left sinus of Valsalva is a rare congenital disease. Malignant course means passing between the main pulmonary artery and the aortic root. It is relatively rare but may present with angina or sudden cardiac death especially in young patients.

Here, we report a case of a male patient 41 years old who complained of persistent chest pain. Further workup demonstrated high-sensitivity troponin that peaked at 910 ng/dl. He was investigated for acute coronary syndrome (ACS), coronary angiography done which revealed an abnormal origin of the RCA arising from the left coronary cusp and left circumflex artery (LCX) originating from proximal RCA with no flow limiting lesions. He underwent a computed tomography (CT) coronary angiography, which revealed a malignant inter-arterial course between the aorta and main pulmonary artery.

Definitive therapy is surgical intervention with unroofing of intramural segments, stenting, or surgical intervention with bypass grafting or translocation of the anomalous artery. However, in older patients, conservative management with exercise limitations is an acceptable option.

Keywords: abnormal origin, inter-arterial course, CT coronary angiography, sudden cardiac death.

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INTRODUCTION

An abnormal origin of coronary artery from the contralateral sinus is a rare condition, even if the patient is asymptomatic, it is an incidental finding during coronary catheterization. 81% were benign anomalies, whereas the rest included abnormal origin from the pulmonary artery, abnormal origin from the contralateral aortic sinus, single coronary artery, and large coronary fistulae with profound consequences (1). This 19% contributes to one-third of sudden cardiac deaths (SCD) in young patients and is the second leading cause of SCD in athletes after hypertrophic cardiomyopathy (2).

In a coronary angiography study of 1,950 patients by Angelini et al., the incidence of anomalous coronary arteries was 5.6%. The incidence of the RCA arising from the left coronary cusp was 0.92%. The ARCA is more prevalent than the anomalous left coronary artery (ALCA) and accounts for a majority of SCD (3). Patients with ALCA are younger in age compared to ARCA. Mortality rates are higher in ALCA (57%) when compared to ARCA (25%) (4).

Case presentation

A 41 years old male diabetic, hypertensive, dyslipidemic presented with recurrent retro sternal chest pain. During examination, his heart rate was 76 bpm and the measured BP was 134/88 mm Hg. The other vital signs were unremarkable. The physical examination revealed normal cardiovascular and respiratory system examination.

The baseline serum creatinine was 1.04 mg/dl and the high sensitivity troponin was 910 ng/dl. A transthoracic echocardiogram revealed an ejection fraction of 55% without any wall motion abnormalities. There was trivial mitral regurgitation and mild tricuspid regurgitation. After giving informed consent, he underwent coronary artery catheterization which revealed no flow limiting coronary artery lesions with abnormal origin of the RCA from the left coronary cusp and LCX arising from RCA (Figures 1). It was suspicious of the probable inter-arterial course of the RCA. CT coronary angiography done which revealed abnormal origin of RCA arising from the left coronary cusp and LCX arising from

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RCA (Figures 2). It demonstrated an inter-arterial course between the main pulmonary artery and the ascending aorta. Decision was taken for surgical intervention.

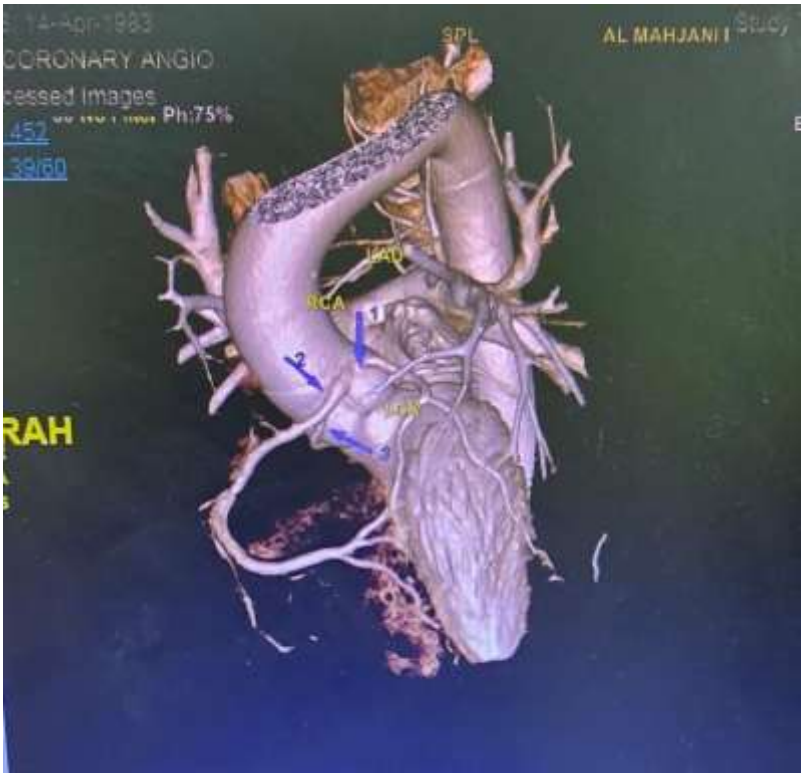

Figure 1: abnormal origin of RCA and LCX from left coronary sinus

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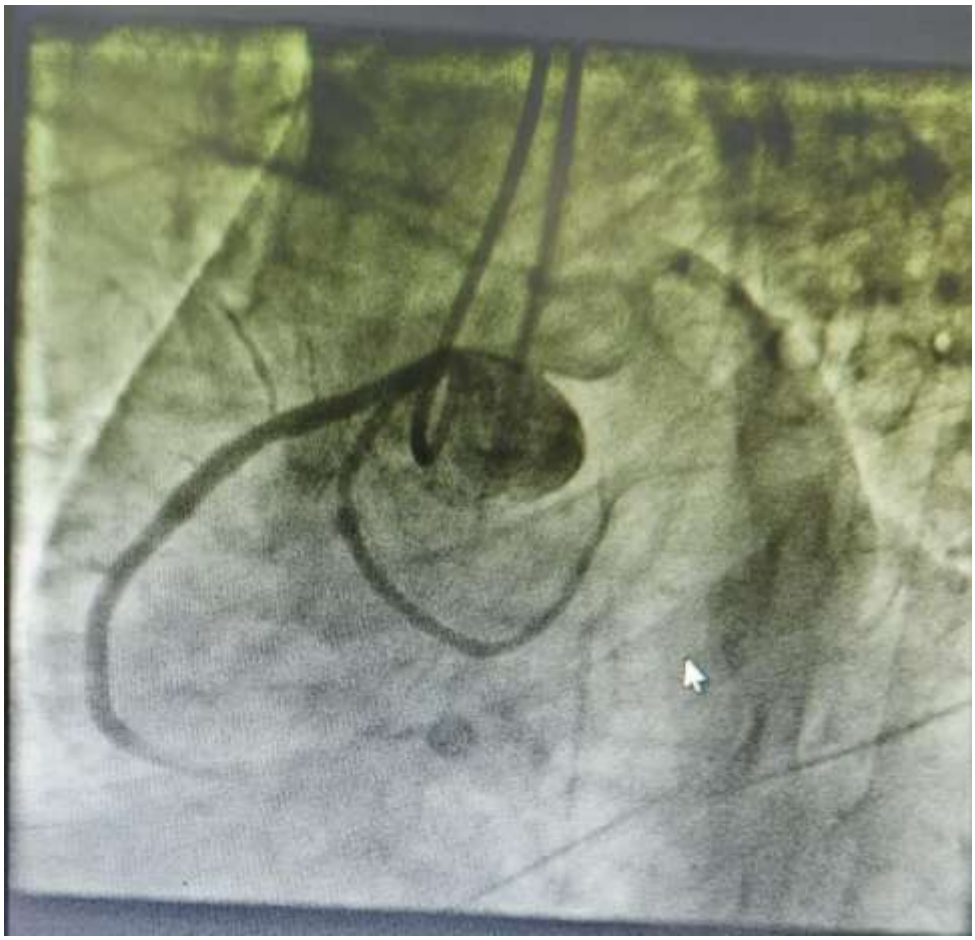

Figure 2: pre-operative coronary angiography

Procedure

Under general anesthesia, median sternotomy, full heparinization, aortocaval cannulation, dissection of abnormal origin of RCA from left coronary cusp, dissection of LCX from RCA, initiation of cardiopulmonary bypass (CPB), translocation of RCA from left coronary cusp to right coronary sinus, smooth weaning from CPB, closure in layers.

Post-operative patient course was smooth, no ECG changes, no chest pain, and CT coronary angiography revealed translocated RCA to right coronary sinus without stenosis. (Figures 3)

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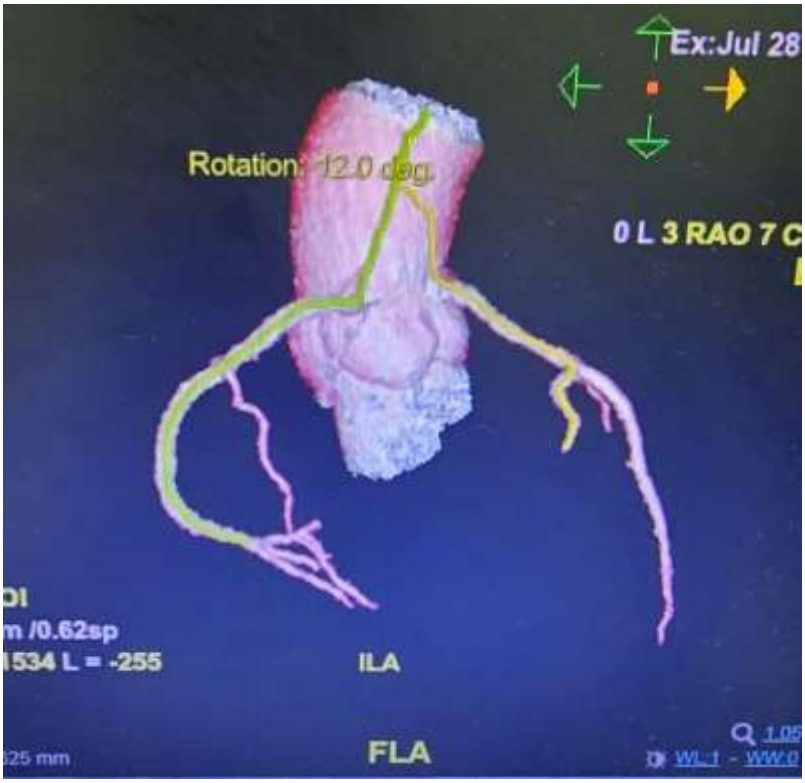

Figure 3: post-operative translocation of RCA and LCX origin

Discussion

A symptomatic anomalous coronary artery from the contralateral sinus presents with symptoms similar to ACS, ARCA is further classified according to its course into two categories: a high inter-arterial course (when ARCA travels between the aorta and pulmonary artery) and a low inter-arterial course (when ARCA travels between the aorta and right ventricular outflow tract). The reason for differentiation is important as the higher course is malignant course and associated with major adverse cardiac events (MACEs) and most likely will need surgical intervention when compared to low course ARCA.(5)

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The pathophysiology based on current theories is transient ischemia. Proposed mechanisms are: (A) ostial stenosis due to acute take-off angle, slitlike orifice, and compression of the intramural segment by the aortic valve commissure, (B) mechanical compression, which is contributed to adrenergic effect during exercise, leads to increased cardiac output resulting in expansion of the aorta and pulmonary artery, which leads to mechanical compression of the RCA, and (C) vasospasm of anomalous artery (6).

An intravascular ultrasound study states that luminal compression of the coronary artery was totally attributed to the aorta because pulmonary artery pressure was lower than the aorta. Some autopsy-based studies state ostial stenosis is associated with sudden cardiac death. This transient ischemia leads to malignant arrhythmias and SCD (6).

Once discovered, the treatment options need to be carefully recognized. In patients with asymptomatic or symptomatic ALCA, surgical repair is indicated due to its substantial risk of sudden cardiac arrest. But in patients with ARCA, the treatment path is not clear as most cases are benign due to the lower risk of SCD in these patients, especially if it is not a higher inter-arterial course(7).

Older patients should most preferably choose conservative management with exercise limitation due to its benign course and conservative treatment, unroofing of intramural segments, stenting or surgical intervention with bypass grafting or re-implantation of the anomalous artery(8).

Coronary angiography and angioplasty with stent placement are difficult due to small, slit-like orifices and long, curved intramural portions of the anomalous artery. Success rates of selective cannulation are 55-61%, which is also because of limited experience by interventional cardiologists due to its rarity (8).

The unroofing procedure enlarges the orifice, creates an acute angulation, and decreases lateral compression of the intramural segment. Complications involve aortic valve incompetence. The coronary artery bypass graft is easier, but the native artery is patent at rest, leading to competition flow, which can be overcome

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by ligation of the native anomalous artery. Coronary reimplantation to the right coronary sinus is possible but has a risk of neo-ostial stenosis (8).

In our case, RCA re-implantation was done with smooth postoperative course with no ECG changes, hemodynamically stable and no cardiac enzymes elevation. Post-operative CT coronary angiography was done which showed patent new origin with normal coronary blood flow. The patient's follow up after three months we found that the patient was free of chest pain with normal ECG.

Conclusion

Although abnormal origin of coronary arteries from the contralateral sinus is uncommon, they can lead to disasters such as SCD The greatest challenge is to detect the abnormality accurately, as cardiologists are unaware of this disease due to its rarity and should keep this as a differential diagnosis. Routine testing with electrocardiography and echocardiography is not sensitive enough to diagnose congenital abnormalities and needs further investigation.

Cardiac surgeons have to take proper decision for surgical intervention according to the history of the patient and the investigations

Human Ethics

Consent was taken from all participants in this study

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