CASE SERIES

SYMPTOMATIC INTERARTERIAL COURSE OF CORONARY ARTERIES IN MIDDLE AGED PATIENTS

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Anomalous aortic origin of a coronary artery (AAOCA) is the most common congenital abnormality and is sometimes associated with various life-threatening conditions. We present the cases of a 35-year-old male and a 50-year-old female with complaints of chest pain. Patients had anomalous aortic origin of coronary arteries with the interarterial course and were treated surgically. By literature review, we came to know that the approach to treat patients with anomalous aortic origin of coronary arteries should be largely individualized and there is no ample scientific data to support any specific diagnostic modality and treatment option.

Keywords: AAOCA; Congenital abnormality; Anomalous aortic origin

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INTRODUCTION

Coronary artery abnormalities e.g., anomalous origins of coronary arteries, myocardial bridging and coronary artery fistula are the most common congenital cardiovascular diseases. Among them, the anomalous origin of the coronary artery is sometimes associated with life-threatening situations. The most fatal is the origin of coronary arteries from pulmonary arteries. Anomalous origin of coronary arteries from inappropriate sinus is also another important cause of SCD. The prevalence of AAOCA in one study in which CTA was used as a diagnostic modality was found to be 1.5%.[1]

Patients with anomalous origins of coronary arteries present with a wide spectrum of symptoms ranging from being completely asymptomatic to having chest pain, arrhythmia and sudden cardiac death. In one autopsy study anomalous origin of coronary arteries was responsible for one-third of cases of SCD among military recruits.[2]

For the appropriate treatment of the anomalous arteries, one has to look for the underlying mechanism. Not only the vessel's origin but its course is also important. The most malignant feature is the interarterial course, i.e., the vessel travelling between great arteries. According to one study the highest incidence of interarterial course occurs with RCA when originating from an inappropriate sinus (0.03% for anomalous LCA and 0.23% for anomalous RCA)[3] and the highest incidence of SCD is noted with anomalous LCA.[2]

Because of the lack of sufficient scientific data on this rare anomaly, there is a great disparity between different practitioners in managing the anomalous aortic origin of coronary arteries. Some believe in surgical correction whether or not ischemia is present, while others in a more conservative approach.

CASE REPORT 1

35 years old male with no previous co-morbid and low-risk profile for ischemic heart disease, presented with a longstanding history of cardiac ischemic pain to our outpatient department. He had been on multiple antianginals with insignificant relief. His resting ECG was normal. The echocardiogram was also unremarkable with normal chamber sizes and function.

The patient had an exercise tolerance test which was highly suggestive of ischemia and it stopped when for patient became symptomatic. The patient was then booked for CCTA which revealed a right coronary artery arising from the left coronary cusp, having a slit-like origin and brief course between the aorta and pulmonary artery before becoming slightly ectatic and running down its usual course (Figure 1(a, b) & 2). For the suspicion of ostial RCA disease and more formal assessment, the patient underwent invasive angiography which showed an ectatic right coronary artery with anomalous origin. (Figure 3).

Based on symptoms and exercise ECG patient underwent surgery with grafting of distal RCA with the right internal mammary artery (RIMA). The patient followed through clinic visits with complete resolution of symptoms.

Figure-1: (a) CT axial plane showing the origin of the right coronary artery (RCA) from the left cusp.

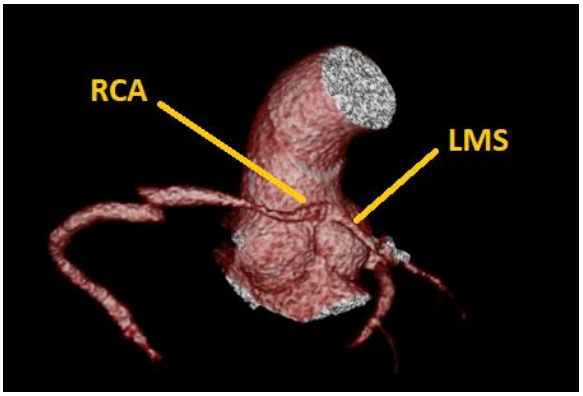

Figure-1: (b) 3D image showing anomalous RCA (right coronary artery) originating from left cusp along with LMS (left main stem).

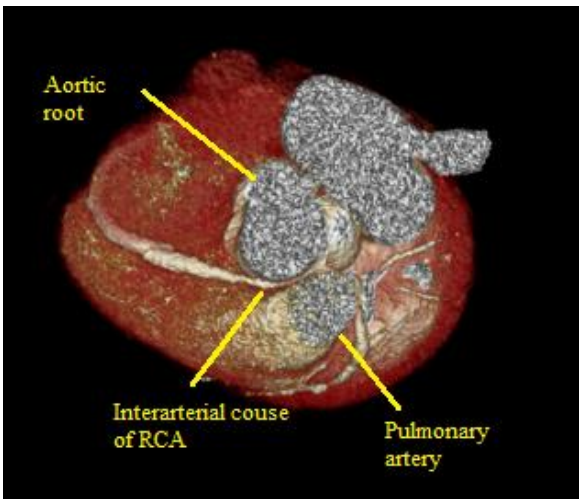

Figure-2: 3D CT image showing interarterial course of right coronary artery (RCA) with proximal narrowing.

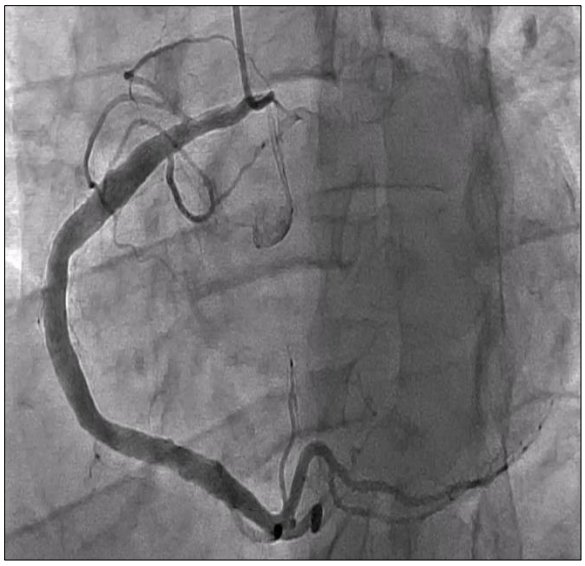

Figure-3: Angiographic views demonstrating the origin of the right coronary artery from the left cusp.

CASE REPORT 2:

A few weeks later another patient presented to our hospital. She is 50 years old with 1-year history of hypertension which was well controlled with beta-blockers. The patient had atypical chest pain for the last 3 years which had no association with exercise. However, it was radiating towards the arms and back. The symptoms increased in frequency and intensity three weeks before presenting to us. ECG showed non-specific T wave inversion in anterior leads. Echo demonstrated good left ventricular function.

She was put on optimum antianginals but reported no relief on a follow-up visit. The patient then underwent an invasive angiogram which revealed an anomalous origin of the left main stem from the right cusp (Figure 4), followed by a CT coronary angiogram to delineate the course of the left coronary artery. It shared a common origin with the right coronary artery from the right cusp, coursing between the pulmonary artery and aorta before bifurcating into the Left anterior descending and left circumflex artery (Figure 5).

Taking into account the high-risk anatomy of the left coronary artery and the associated risk of sudden cardiac death, she was offered surgery. It was an off-pump CABG and LAD was grafted in the mid-course with a long saphenous vein. The patient reported complete resolution of symptoms on follow-up in the clinic.

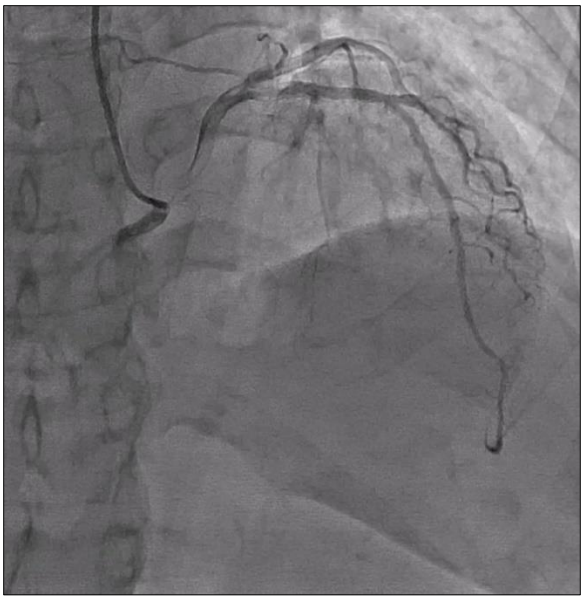

Figure-4: coronary angiogram demonstrating the origin of the left main stem from the right cusp.

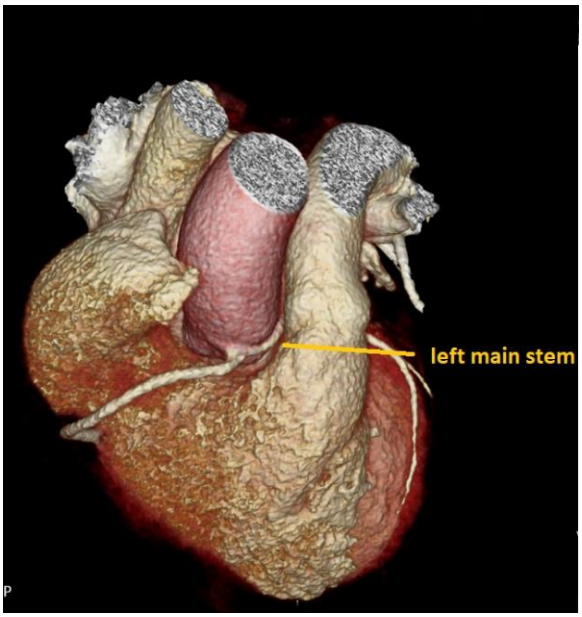

Figure-5: 3D CT image showing the origin of the left main stem from the right cusp and its interarterial course.

DISCUSSION

Anomalous aortic origin of coronary artery (AAOCA) accounts for the second highest cause of SCD after HOCM.[4] Various mechanisms and their complex interplay have been postulated for myocardial ischemia in patients with anomalous origins of the coronary arteries besides the interarterial course. Considering the low pressures in the pulmonary artery, it is less likely that coronary arteries can get compressed between great vessels (however may be possible when there is pulmonary hypertension). The interarterial course itself is not malignant but acts as an indicator of other high-risk features. Those include acute origin, slit-like ostium, coronary spasm, elliptical vessel and intramural course, i.e., vessel traversing within the tunica media of the aortic wall.[5]

2018 ACC guidelines of adult congenital heart disease recommend surgical correction of AAOCA (anomalous coronary arteries arising from opposite sinus) if the patient has ischemic symptoms or demonstrates ischemia during stress testing (Class 1, Level B-NR). Surgery or close surveillance may be reasonable for asymptomatic patients with suitable anatomic features, but the level of recommendations is weak due to the low incidence of the disease.

The modality which tends to be used for the detection of anomalous coronary arteries should be able to define the associated pathological mechanism and its functional significance. This is not possible with a single modality. Due to the incompletely understood pathophysiology of the anomalous origins of the coronary arteries and its implication in causing myocardial ischemia, various diagnostic modalities currently available for the detection of the physiological significance of coronary artery disease are not applicable for detecting the hemodynamic significance of the anomalous coronary arteries.

CCTA has been the established modality for the identification of the origin of coronary arteries and their high-risk features. MRI is another modality which can overcome hazards of radiation and contrast but at the cost of low resolution. CCTA can successfully delineate the interarterial course of arteries but identification of the intramural segment will be difficult. However, the presence of slit-like ostium and proximal narrowing are the features which correspond to the intramural course of coronary arteries from ectopic origin.[6] IVUS is the most useful modality in detecting the intramural course of coronary arteries.[7]

Ischemia testing can be done by non-invasive or invasive methods. Various experts suggest that the absence of ischemia during functional testing should not be relied upon especially when associated with high-risk features and patients with high-risk features should be treated surgically.[3]

However Japanese experts believe in more conservative management of such groups of patients. One Japanese study comprising 56 patients with anomalous origin of the right coronary arteries from the left aortic cusp demonstrated successful management of such patients with exercise restriction and the use of beta blockers.[8] However our patients were symptomatic despite medical treatment so further continuation of medical treatment without

surgical correction was not deemed a good idea. Also, the study included patients exclusively having anomalous RCA.

Various surgical treatments have been proposed which are employed according to expertise availability, i.e., surgical unroofing (usually done when AAOCA is associated with intramural course), osteoplasty (creating a neo-ostium for the artery from its respective sinus) and bypass grafting with proximal coronary artery occlusion (coronary artery occlusion is necessary because the competitive flow from the relatively patent artery will be a threat to patency of the graft). The choice and success of surgical technique depends highly on the expertise available. Mostly paediatric surgeons are considered more expert in osteoplasty, however the patients with anomalous origin usually present in adulthood.

Although surgery is the treatment of choice recently percutaneous coronary interventions have shown promising results, especially in patients with an interarterial course of the right coronary artery.[9]

Still, no long-term data is available for the prognosis of such patients if they had ever undergone surgical correction (or which surgical technique has got better outcome) or managed with a more conservative approach.

Conflict of interest:

All authors hereby declare no conflict of interest.

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