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Case Report

Shepherd's crook configuration of right coronary artery and anomalous origin of left circumflex artery from right coronary cusp in a middle-aged man—A case report ☆

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ABSTRACT

Shepherd's crook configuration of the right coronary artery is a course anomaly where the ostium is oriented superiorly with the proximal artery taking an upward turn before resuming its regular path. Although it is classified as an unimportant hemodynamic variation, it is relevant in the context of coronary artery disease due to the technical issues it causes when being treated. The anomalous origin of the left circumflex artery arising as a separate branch from the right coronary cusp is a rare variant and its significance lies in its association with sudden arrhythmia, syncope, and sudden cardiac death. Here we report a case of a 58-year-old male patient with an anomalous course of the right coronary artery consistent with Shepherd's crook configuration and anomalous origin of the left circumflex artery from the right coronary cusp.

Introduction

The right coronary artery emerges from its ostium in the right sinus of Valsalva, which is found between the aortic valve annulus and the sinotubular junction. The proximal section of the right coronary artery travels superiorly and rightward, posterior to the pulmonary trunk, and below the right atrial appendage following a sharply horizontal egress from the aorta. In the atrioventricular groove's plane, the mid-right coronary artery turns inferiorly. The distal segment proceeds along the posterior interventricular groove and turns toward

Abbreviations: Cx, Circumflex artery; ECG, Electrocardiogram; LCx, Left circumflex artery; PDA, Posterior descending artery; PLV, Posterior left ventricular artery; RCA, Right coronary artery.

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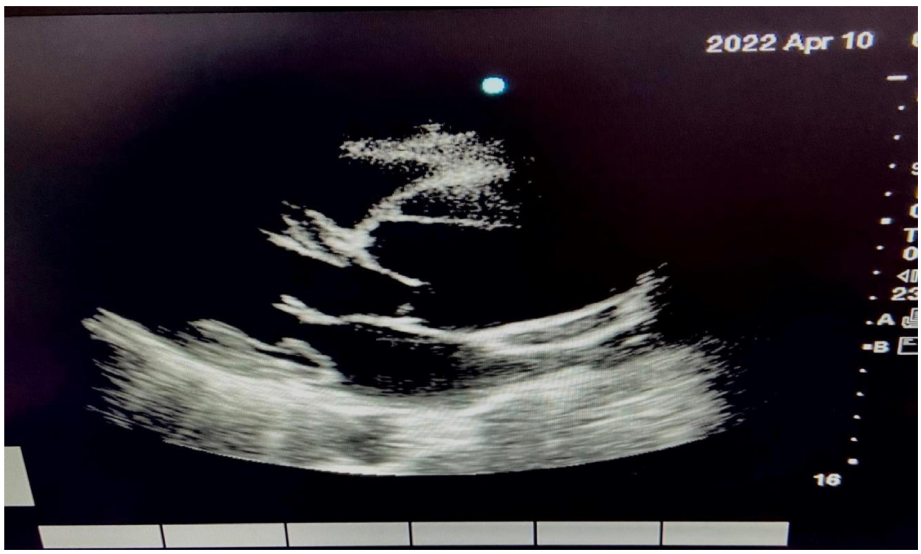

Fig. 1 – Parasternal long axis view 2D Echocardiography image showing normal left atrium, left ventricle, interventricular septum and posterior wall. Left ventricular ejection fraction was 55% (not shown).

Fig. 2 – Volume rendered images show superiorly directed right coronary artery (RCA) ostium with an upward curve followed by a U-turn to travel caudally along its expected course supplying the right ventricular myocardium (A-C). Anomalous origin of left circumflex artery from right coronary cusp via separate ostium (white arrow). Normal origin of left coronary artery from left coronary cusp (D).

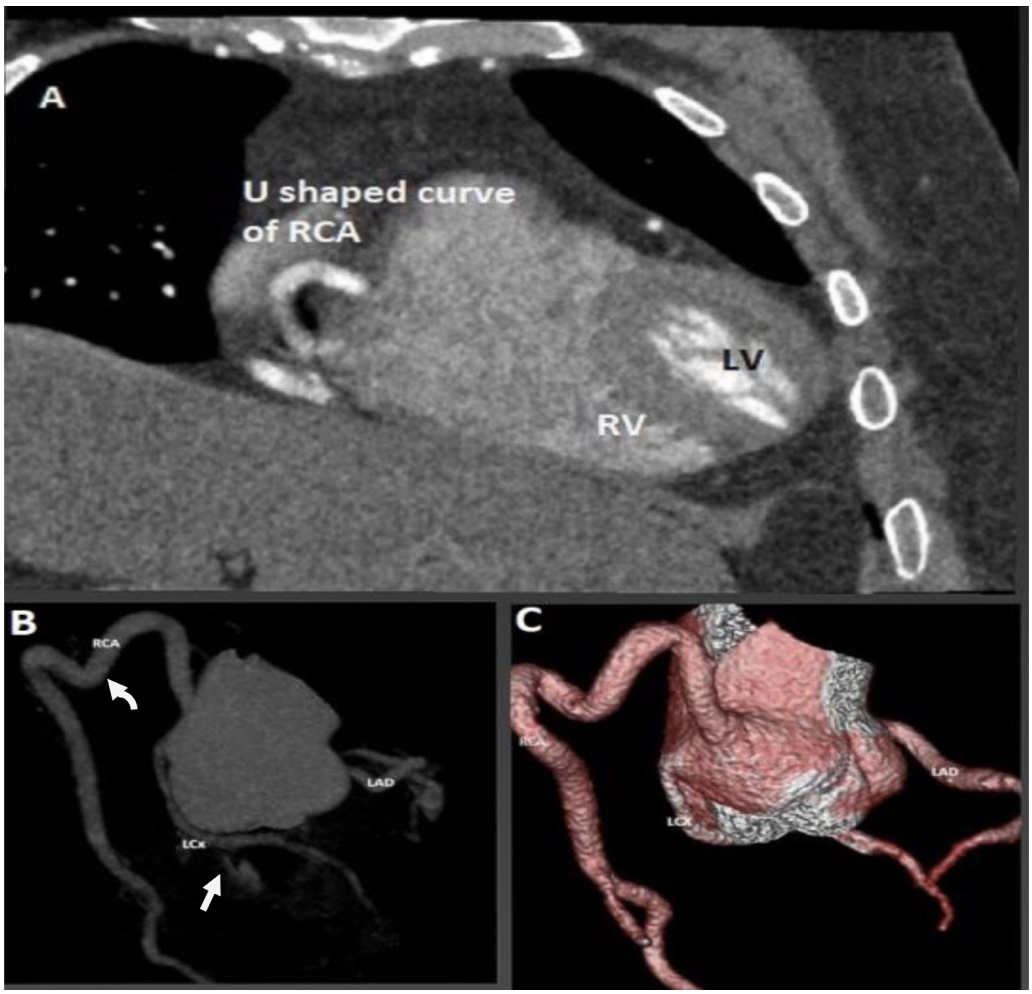

Fig. 3 – (A) CT coronary angiography image in coronal plane showing U shaped curve of right coronary artery. (B) CT coronary angiography image showing the origin of RCA from right coronary cusp with Shepherd's crook configuration (curved arrow in B). There is anomalous origin of LCx from right coronary cusp via separate ostium (straight white arrow in B), (C) volume rendered image showing RCA and LCx arising from right coronary cusp via separate ostium.

the cardiac crux before bifurcating to the posterior left ventricular (PLV) branch and the posterior descending artery (PDA) [1]. Normally, the RCA does not bend as it travels through the atrioventricular groove [2]. Shepherd's crook configuration often involves main RCA in which the ostium of RCA is directed superiorly and the proximal RCA courses upwards before making a U-turn to enter the right atrioventricular groove [3]. This configuration is regarded as an abnormality of course that is not significant hemodynamically. However, Shepherds' crook artery requires extra attention since it interferes with right coronary artery procedures [2].

One of the 2 primary coronary arteries that emerge from the bifurcation of the left main coronary artery is the circumflex artery (Cx), with the left anterior descending (LAD) artery serving as the other branch. The artery runs between the left atrium and left ventricle in the left atrioventricular groove. The circumflex artery gives rise to 3 obtuse marginal branches and may give off a left posterolateral branch and may supply the inferior interventricular artery [4]. It is thought to be a rare variation where the left circumflex artery (LCx) develops abnormally as a separate branch from the right coronary cusp. The anomalous LCx passes posterior to the aortic root through the atrioventricular groove to supply the lateral wall of the left ventricle. Although this anomalous origin typically has a benign and asymptomatic course, its relationship with sudden cardiac death, syncope, and arrhythmias as a sign of myocardial ischemia makes it clinically significant [5]. Some have suggested that the retro aortic course and acute angulation of its aortic origin may lead to an increase in coronary artery disease in these arteries [6].

Case presentation

We present a case of a 58-year-old male patient who presented in the outpatient department of our hospital with a 2-month history of exertional chest pain and dyspnea. He was then reviewed with echocardiography which revealed normal findings with an ejection fraction of 55%.

Electrocardiogram showed minimal T wave changes in the inferior leads. Troponin T was normal. He was then advised for CT coronary angiography. The review of the multiplanar reconstructed and volume-rendered CT angiography images showed right dominant circulation without any substantial coronary artery disease. Incidentally, an anomalous origin of LCx was noted arising from the right coronary cusp. The anomalous LCx passed posterior to the aortic root through the atrioventricular groove to supply the lateral wall of the left ventricle. In addition, Shepherd's crook configuration of the right coronary artery was identified with the artery having a tortuous and high course, just after its origin from the aorta.

The past medical history of the patient was significant for hypertension for which he has been taking amlodipine and losartan. Examination showed blood pressure of 150/90 mm Hg, Pulse of 92 bpm, Temperature of 98 degree F, and Respiratory rate of 18 breaths per minute. His physical examinations were unremarkable. Baseline hematological tests revealed normal findings.

Discussion

A coronary artery with a shepherd's crook configuration often has a superiorly orientated ostium and a proximal artery that ascends before turning around and continues along its normal path. The most common coronary artery to present with this configuration is right coronary artery (RCA) with an estimated prevalence 5% [3].

Even though it is not clinically significant, it carries relevance in the context of coronary artery disease due to the numerous technical issues it can cause during the course of treatment. The difficulty in steering the guidewire and balloon over the lesion is one of the procedural obstacles. This difficulty is linked to a lower primary success rate for percutaneous coronary interventions and a higher frequency of acute complications, such as dissections and abrupt occlusions [7]. Low success rate during catheterization and increased incidence of procedural difficulties were found in a study by Gossman et al. with this anomalous configuration of the coronary artery [8]. Special catheters are required for an abnormal course of the right coronary artery in order to prevent difficulties during the surgery [2].

The anomalous origin of the LCx from the right coronary cusp is a rare entity. Antopol and Kugel first identified the anomalous origin of the LCx from the right coronary system in 1933 and have an estimated frequency of 0.32%-0.67% [5].

The anomaly can be classified based on the site of origin into various subtypes [5].

- The LCx directly branches off from the right coronary artery,
- A common right system ostium bifurcating into the LCx and RCA and,
- The LCx and the RCA emerge from 2 different orifices as in our case.

Complications of the anomalous origin and course of LCx from the right coronary cusp can arise due to the slit-like ostia, the vessel's deviation off its retro aortic path, or the compression of the anomalous artery by dilated aorta which in turn can lead to myocardial ischemia and sudden cardiac death [6].

Conclusion

Shepherd's crook configuration of RCA is a course anomaly that has significance as it may pose procedural challenges during the coronary intervention and may result in lower primary success rates and higher complication rates. Anomalous origin of the LCx from the right coronary system is a relatively rare anomaly but significant as it may give rise to complications like myocardial ischemia, syncope, and arrhythmia (Figs. 1-3).

Patient consent

Consent from the patient was taken on written form for the case report and using echocardiography and CT angiography images in any journal after explaining in his own language.

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