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Coexistence of Anomalous Right Coronary Artery Originating from the Left Coronary Cusp and Interventricular Membranous Septal Aneurysm: A Rare Dual Congenital Findings

Khalid Bin Thani[1,2*], Wafa Alhayki[1] and Yousef M. Alajmi[2]

[1]Department of Cardiology, Salmaniyah Medical Complex, Manama, Kingdom of Bahrain

[2]College of Medicine and Medical Sciences, Arabian Gulf University, Manama, Kingdom of Bahrain

[*]**Corresponding author:** Khalid Bin Thani, Department of Cardiology, Salmaniyah Medical Complex, Manama, Kingdom of Bahrain; College of Medicine and Medical Sciences, Arabian Gulf University, Manama, Kingdom of Bahrain.
E-mail: kbinthani@hotmail.com

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Abstract

We present a rare case of a 67-year-old woman with exertional chest discomfort who was found on coronary computed tomography angiography (CCTA) to have two coexisting congenital cardiac anomalies; an anomalous right coronary artery (RCA) originating from the left coronary cusp (LCC) with an inter-arterial course between the aorta and pulmonary trunk, and an interventricular membranous septal aneurysm (IVMSA). Anomalous RCA with inter-arterial course is considered a malignant variant due to its association with myocardial ischemia and sudden cardiac death [1,2,6], while IVMSA, although often incidental, may be associated with conduction disturbances and thromboembolic complications [4,5,8]. The patient was discussed in a multidisciplinary team and surgical correction was offered, but she opted for conservative management and declined surgical intervention. Recognition of such dual congenital anomalies is crucial, as both may carry significant prognostic and management implications. This case underscores the essential role of CCTA in accurately delineating complex coronary and structural anomalies, guiding individualized treatment decisions, and optimizing patient counseling and follow-up [7].

Keywords: Anomalous right coronary artery; Left coronary cusp; Inter-arterial course; Interventricular membranous septal aneurysm; Coronary CT angiography

Introduction

Congenital anomalies of the coronary arteries are uncommon, with an estimated prevalence ranging from 0.2% to 1.3% in the general population, yet they can have significant clinical consequences. Among these, an anomalous origin of the right coronary artery (RCA) from the left coronary cusp (LCC) with an inter-arterial course between the aorta and pulmonary trunk is of particular clinical relevance due to its association with myocardial ischemia, arrhythmia, and, in some cases, sudden cardiac death, even in the absence of atherosclerotic disease [1,2]. The underlying mechanisms include slit-like ostial morphology, acute angulation of the origin, and potential dynamic compression of the artery between the great vessels during exertion [3]. An interventricular membranous septal aneurysm (IVMSA) is another rare congenital finding, typically identified incidentally during imaging or autopsy. While often benign and hemodynamically insignificant, IVMSA may be associated with conduction abnormalities, thromboembolism, or coexistent structural cardiac defects [4,5]. The coexistence of an anomalous RCA arising from the LCC with an inter-arterial course and an IVMSA in the same patient is exceedingly rare, with only a few cases reported in the literature. Recognition of such combined anomalies is important for comprehensive diagnostic evaluation, risk assessment, and management planning. Herein, we report the case of an elderly woman presenting with exertional chest heaviness who was found to have both anomalies on CCTA. This case highlights the critical role of advanced cardiac imaging in the identification of rare congenital abnormalities in adults and discusses their potential clinical implications and management strategies considering the existing literature.

Case Presentation

A 67-year-old female with a medical history of hypertension, bronchial asthma, dyslipidemia, and long-term heavy smoking presented with intermittent, non-radiating chest heaviness exacerbated by exertion and associated with shortness of breath. On examination, she was hemodynamically stable with a mild elevation in jugular venous pressure; other findings were unremarkable. A baseline electrocardiogram (ECG) (Figure 1) demonstrated normal sinus rhythm, left ventricular hypertrophy (LVH) with strain pattern and right bundle branch block (RBBB). The transthoracic echocardiography revealed preserved left ventricular systolic function with an ejection fraction greater than 55%, mild concentric left ventricular hypertrophy, and a thinned, aneurysmal basal interventricular septum located just below the aortic valve, without evidence of a shunt by color-Doppler or by bubble-contrast study (Figure 2A-C), and a normal right ventricular systolic pressure. Therefore, a Coronary CT angiography (CCTA) was performed showing no evidence of a significant coronary artery atherosclerosis with a moderate coronary artery calcification. The right Coronary Artery (RCA) is the dominant vessel with anomalous origin from the left coronary cusp (LCC). The vessel followed an inter-arterial course between the ascending aorta and a dilated main pulmonary artery consistent with pulmonary hypertension, and the inter-arterial coronary course is suggestive of mechanical compression (malignant course). The proximal RCA showed luminal narrowing related to its aberrant course. The interventricular membranous septal is aneurysmal (IVMSA) with an outpouching of the membranous septum projecting into the right ventricle, partly formed by the septal leaflet of the tricuspid valve; with no shunt or ventricular septal defect (VSD) identified (Figure 3-5). Of other significant findings there was a peripheral pulmonary vasculature pruning and distal vessel loss, suggestive of chronic pulmonary hypertension.

In summary, the patient findings are anomalous RCA originating from the LCC with inter-arterial course (malignant variant) with an interventricular membranous septal aneurysm (IVMSA) and associated with pulmonary artery dilation, right heart enlargement, and moderate coronary calcification.

The patient condition and findings were discussed in a multidisciplinary team, and she was offered surgical correction. But the patient preferred conservative management with medical therapy with a close follow-up visit.

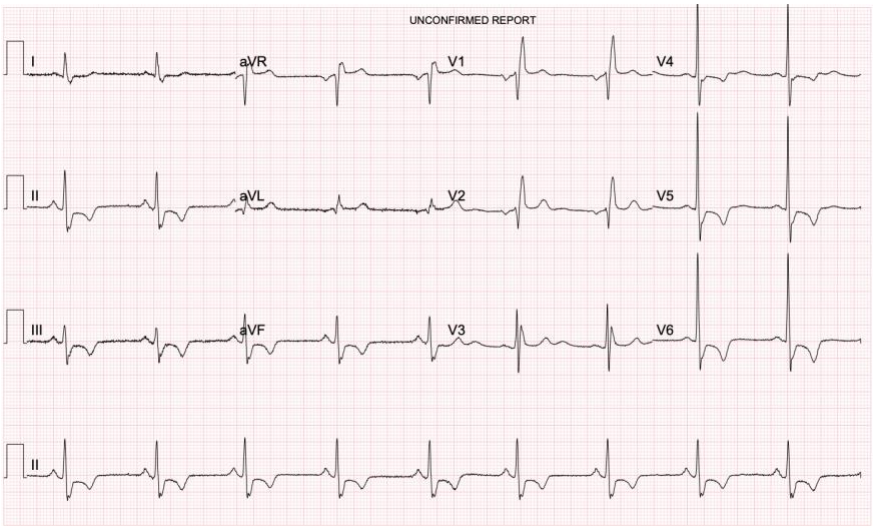
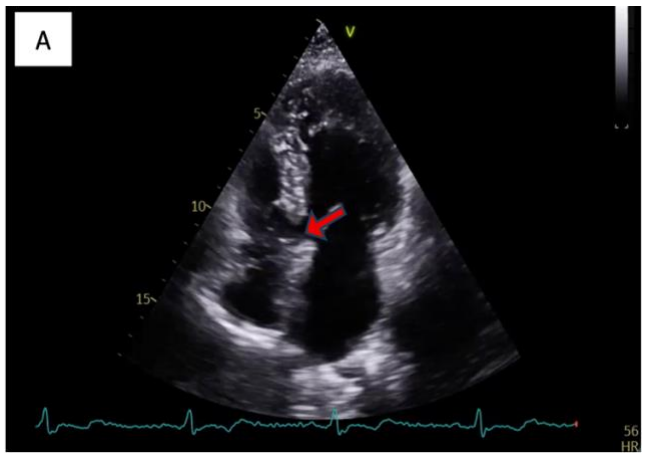
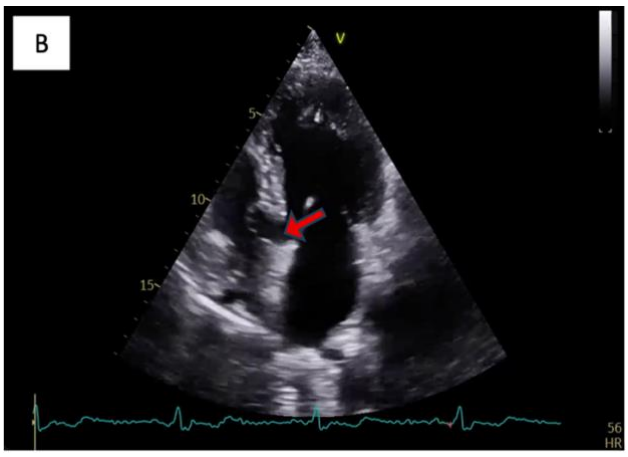


Figure 1: ECG: sinus rhythm, LVH with strain pattern "downsloping ST depression and T-wave inversion in lateral leads", RBBB.





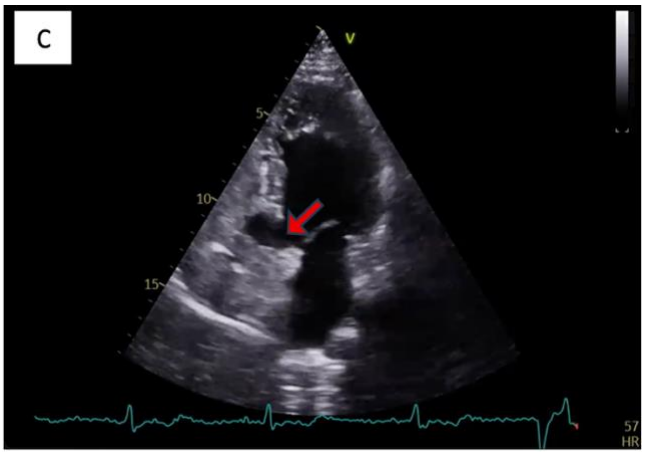


Figure 2(A-C): Apical four-chamber transthoracic echocardiographic views demonstrating an aneurysmal outpouching of the basal interventricular septum (red arrows), consistent with an interventricular membranous septal aneurysm (IVMSA) protruding into the right ventricular cavity. The aneurysmal segment is best visualized in mid-systole and persists throughout the cardiac cycle. (C): Bubble-contrast study shows no evidence of intracardiac shunt, excluding an associated ventricular septal defect.

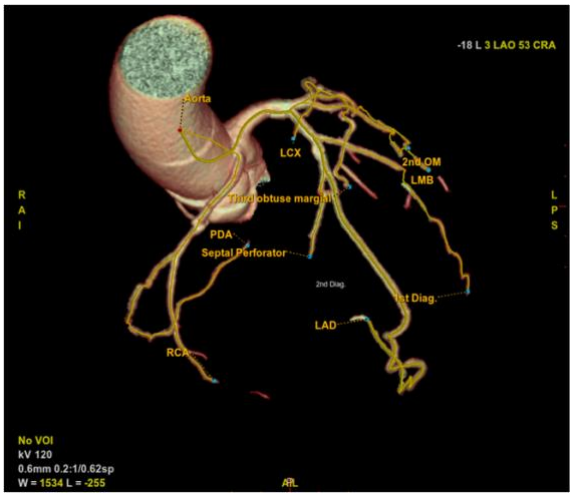


Figure 3: Three-dimensional volume-rendered CCTA showing an anomalous right coronary artery (RCA) arising from the left coronary cusp (LCC) and following an inter-arterial course between the ascending aorta and main pulmonary artery (malignant variant). The left coronary system (Left anterior descending coronary artery [LAD] and left circumflex artery [LCx]) are normal.

Figure 4: The axial CCTA image showing interventricular membranous septal aneurysm (IVMSA) [arrow] projecting into right ventricle without shunt.

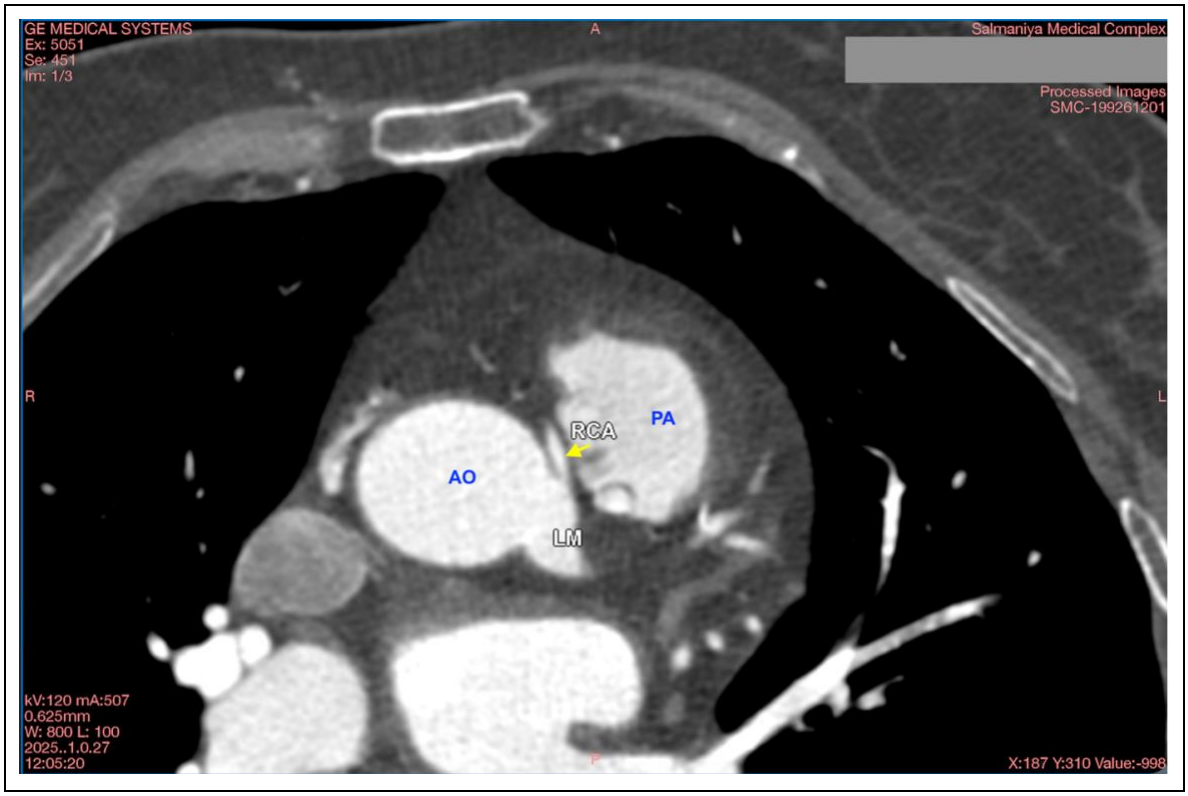


Figure 5: Axial contrast-enhanced coronary CT angiography image at the level of the great vessels demonstrates an anomalous right coronary artery (RCA) arising from the left coronary cusp, with the proximal segment (yellow arrow) coursing between the ascending aorta (AO) and the main pulmonary artery (PA), consistent with an inter-arterial (malignant) course. The proximal RCA appears narrowed and slit-like, likely related to extrinsic compression between the great vessels and acute angulation at its origin. The left main coronary artery (LM) arises normally from the left coronary cusp. This configuration is associated with increased risk of myocardial ischemia and sudden cardiac death, particularly during exertion.

Discussion

Anomalous coronary artery origin is a rare congenital finding but can have important clinical consequences, particularly when the artery courses between the great vessels. The anomalous RCA arising from the LCC with an inter-arterial course is classified as a malignant variant due to the risk of compression between the aorta and pulmonary trunk during exertion, potentially resulting in ischemia or sudden cardiac death [1,6]. The mechanisms include acute angulation, intramural segments, and slit-like ostia predisposing to flow limitation [3]. Non-invasive imaging modalities such as CCTA and cardiac MRI have become essential for accurate anatomical delineation and risk stratification [7]. CCTA provides excellent spatial resolution for identifying the vessel origin, course, and relationship to surrounding structures. In this case, it clearly demonstrated an inter-arterial malignant course with proximal narrowing, highlighting the diagnostic strength of advanced imaging.

The interventricular membranous septal aneurysm (IVMSA) is an uncommon structural anomaly, often discovered incidentally. Although frequently benign, IVMSA has been associated with conduction disturbances, thromboembolic events, and rarely right ventricular outflow tract obstruction. In addition, the aneurysmal membranous septum may incorporate or distort adjacent valvular structures, particularly the septal leaflet of the tricuspid valve and, less commonly, the aortic valve cusps, potentially leading to tricuspid or aortic regurgitation and contributing to progressive ventricular volume overload. Multimodality imaging, including echocardiography and CCTA, is valuable for distinguishing IVMSA from other septal or valvular pathologies [4,5,8].

The coexistence of anomalous RCA originating from LCC with an inter-arterial course and IVMSA is exceedingly rare, with only a few cases documented in the literature. The combination underscores potential developmental correlations in the embryologic formation of the aortic root and membranous septum, although this remains speculative. The dual presence of these anomalies complicates risk assessment, particularly when both may contribute to ischemia, arrhythmia, or structural remodeling.

In this case, CCTA demonstrated dilatation of the main and branch pulmonary arteries, peripheral vascular pruning, and right-sided chamber enlargement, imaging features consistent with pulmonary hypertension. Although interventricular membranous septal aneurysm (IVMSA) is not typically associated with pulmonary hypertension in the absence of a significant intracardiac shunt, pulmonary hypertension may coexist in patients with congenital cardiac anomalies due to long-standing cardiopulmonary disease or prior altered hemodynamics. Given the presence of chronic respiratory disease, including long-standing heavy smoking and bronchial asthma, the pulmonary hypertension in this patient is most consistent with lung disease–associated pulmonary hypertension (World Health Organization Group 3), resulting from chronic hypoxia and pulmonary vascular remodeling [10, 11]. Thus, pulmonary hypertension in this case likely represents a coexisting comorbidity contributing to symptoms and prognosis rather than a direct consequence of the interventricular membranous septal aneurysm.

Management depends on clinical presentation and risk profile. Surgical reimplantation or unroofing of the anomalous RCA is indicated for symptomatic patients or those with demonstrable ischemia. However, in older individuals with comorbidities, conservative management is often reasonable, particularly when symptoms are mild and surgical risk is prohibitive [2,9]. In this case, given the patient's age, comorbidities, and preference, conservative treatment with close follow-up was opted.

Conclusion

This case highlights an exceedingly rare coexistence of an anomalous right coronary artery (RCA) arising from the left coronary cusp (LCC) with an inter-arterial (malignant) course and an interventricular membranous septal aneurysm (IVMSA) in an elderly patient. Recognition of these anomalies is clinically important, as their combination may have additive hemodynamic and prognostic implications, including increased risks of myocardial ischemia and arrhythmia, as well as coexistence of pulmonary hypertension [1,6,10]. Coronary computed tomography angiography (CCTA) plays a pivotal role in diagnosis by providing comprehensive anatomical assessment that guides management decisions [7]. Conservative management was adopted in this case due to patient preference and elevated surgical risk, which is consistent with current recommendations in selected adult patients with anomalous coronary arteries [2,9]. Continued surveillance is essential to monitor for ischemic or hemodynamic complications. Reporting such rare dual congenital findings enriches the literature and underscores the importance of advanced imaging in the evaluation of chest pain in adults.

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