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Case Report

Radiology Section

Anomalous Origin of the Right Coronary Artery from the Pulmonary Artery in a Child with Recurrent Giddiness: A Case Report

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ABSTRACT

Anomalous Origin of the Right Coronary Artery from the Pulmonary Artery (ARCAPA), or abnormal origin of the Right Coronary Artery (RCA) from the main pulmonary artery, is an uncommon congenital coronary anomaly, occurring in fewer than 0.1% of all congenital heart defects and significantly less frequent than Anomalous Origin of the Left Coronary Artery from the Pulmonary Artery (ALCAPA). Myocardial infarction and sudden cardiac death are potential complications, even in asymptomatic individuals. The authors hereby, present a case of a 12-year-old boy who presented with recurrent episodes of giddiness, particularly during prolonged standing. Echocardiography and Coronary Computed Tomography Angiography (CCTA) confirmed the diagnosis of ARCAPA, demonstrating an anomalous origin of the RCA from the Main Pulmonary Artery (MPA) along with mild dilatation of the left atrium and left ventricle. The patient underwent successful surgical repair involving translocation of the right coronary artery from the main pulmonary artery to the aorta. The postoperative outcome was excellent, with stable haemodynamics.

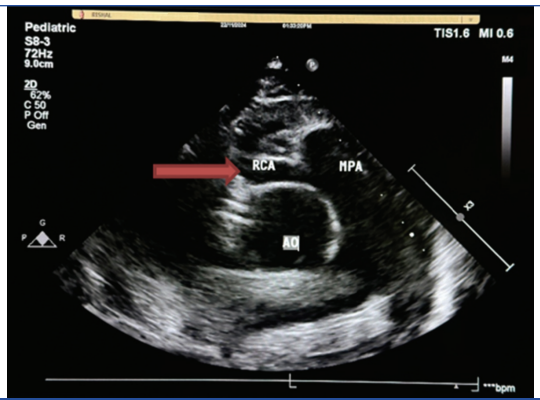
Keywords: Adolescents, Computed tomography angiography, Coronary anomalies, Echocardiography

CASE REPORT

A 12-year-old boy presented with recurrent episodes of giddiness for one year, occurring twice a month, especially during prolonged standing. He had occasional episodes of giddiness since the age of 5 years. He also experienced mild exertional intolerance and dyspnoea on exertion; otherwise, he was normal, with no similar family history. There was no history of failure to thrive or ischaemic chest pain.

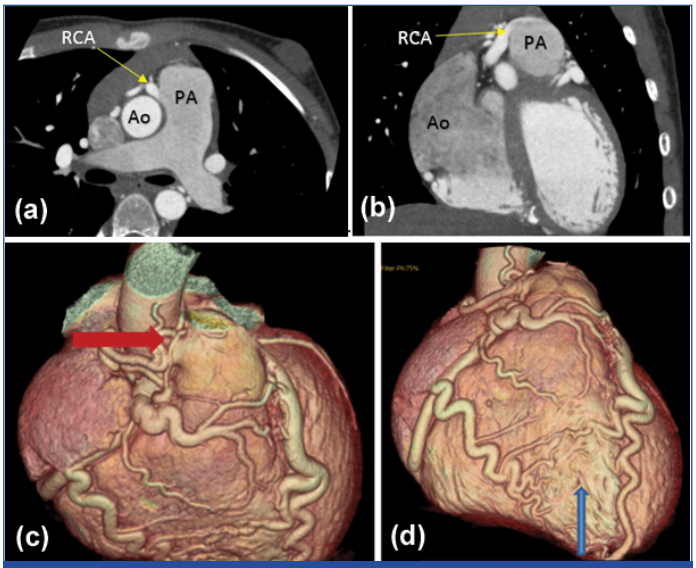
Physical examination revealed a heart rate of 77 beats per minute, Oxygen Saturation (SpO_2) of 100%, and blood pressure of 93/53 mmHg. Cardiac auscultation revealed a short systolic murmur at the left lower sternal border with mild arrhythmia. Chest radiography was normal, and electrocardiography showed no evidence of ischaemia. Routine laboratory results were unremarkable.

An Magnetic Resonance Imaging (MRI) of the brain was performed to rule out causes of dizziness, such as inner ear disorders or cerebellar pathology, but findings were normal. Transthoracic echocardiography showed situs solitus, atrioventricular and ventriculo-arterial concordance, and mild left atrial and left ventricular enlargement. The origin of the RCA from the right coronary cusp was not visualised; instead, an anomalous origin from the MPA was detected [Table/Fig-1].



[Table/Fig-1]: Echocardiography : Parasternal short-axis view at the aortic valve level. Red block arrow: origin of right coronary artery from the pulmonary artery.

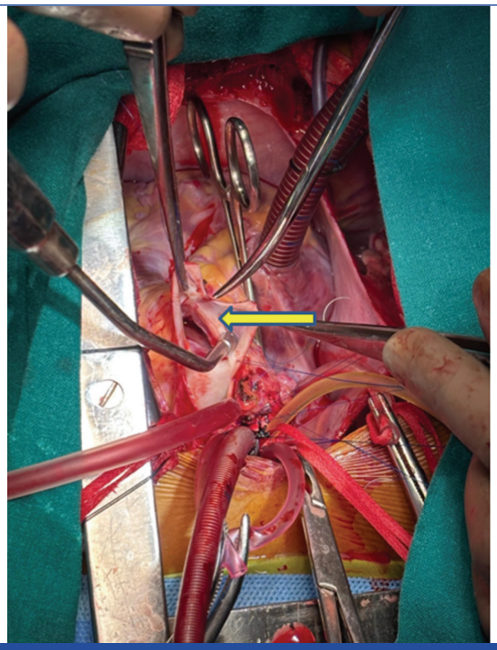
The CCTA confirmed these findings, demonstrating the RCA arising from the MPA, along with mild dilatation of the left atrium and left ventricle [Table/Fig-2a-d]. The patient underwent coronary angiography followed by surgical repair.



[Table/Fig-2]: Coronary CT angiogram in a 12-year-old boy with anomalous origin of right coronary artery from pulmonary artery Ao=Aorta, PA=Pulmonary artery, RCA=Right coronary artery. (a) Axial image showing contrast material flowing retrogradely into the pulmonary artery. This creates a coronary steal phenomenon and is a small left-to-right shunt (Yellow arrow). (b) Coronal image showing the right coronary artery emerging from main pulmonary artery. (c) CT Volume rendering shows an anomalous origin of the right coronary artery from the pulmonary artery (Red Arrow). (d) Rich collateral circulation (Blue arrows) between the Left Coronary Artery (LCA) and Right Coronary Artery (RCA).

Intraoperative findings included situs solitus, levocardia, normally related great arteries, dilated pulmonary arteries, dilated and ectatic coronary arteries, and the RCA originating from the anterior-facing sinus of the pulmonary artery [Table/Fig-3]. Extensive collateral vessels were observed on the epicardial and epiaortic surfaces.

The MPA was transected above the ARCAPA origin, preserving the posterior wall. The RCA ostium was harvested and mobilised. The pulmonary artery was reconstructed using an autologous tanned pericardial patch. The aorta was opened horizontally, stay sutures were placed on the commissures, and the RCA was implanted into the aorta using 6-0 prolene sutures. The aortotomy was then closed.



[Table/Fig-3]: Surgical image showing an anomalous origin of the right coronary artery from the pulmonary artery (Yellow block arrow).

The postoperative period was uneventful. Postoperative echocardiography showed good flow across the translocated RCA, dilated coronaries, unobstructed flow across the MPA, and trivial aortic and mitral regurgitation. The patient was discharged in a stable haemodynamic condition and with a sinus rhythm.

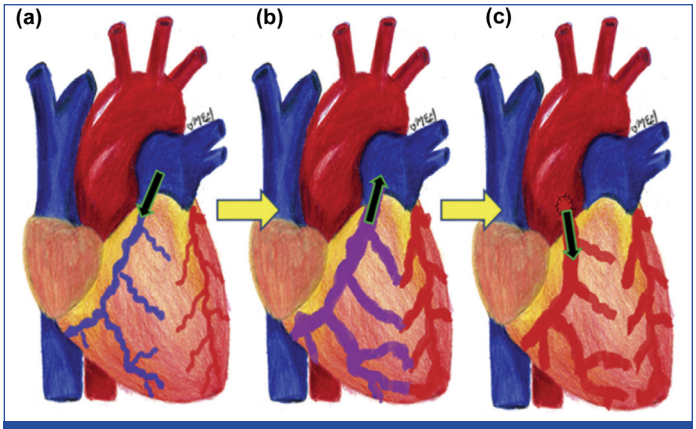
DISCUSSION

The rare congenital coronary anomaly known as ARCAPA can manifest clinically as anything from an asymptomatic murmur to Myocardial Infarction (MI) or sudden cardiac death [1]. Associated congenital heart defects may include aortopulmonary window, ventricular septal defect, atrial septal defect, tetralogy of Fallot, patent ductus arteriosus, aberrant subclavian artery, patent foramen ovale, and bicuspid aortic valve [1]. Its incidence in patients undergoing CCTA is approximately 0.002% [2].

Normally, the RCA and Left Coronary Artery (LCA) originate from the right and left aortic sinuses of Valsalva, respectively. Anomalous coronary artery origins, particularly from the contralateral sinus of Valsalva, are more common than origins from the pulmonary artery. The embryological basis for ARCAPA is likely related to malformations during coronary artery development, which occurs between the fourth and sixth weeks of gestation. Coronary buds appear after the division of the truncus arteriosus by spiral septation into the aorta and pulmonary artery around the 12th day of life. Malpositioning of coronary buds or malrotation of the spiral septum can lead to coronary artery anomalies [3].

The pathophysiology of ARCAPA depends on the direction of blood flow, its impact on myocardial oxygen delivery, and the dominance of the condition [3]. High pulmonary pressure causes antegrade flow of deoxygenated blood through the RCA in patients with ARCAPA from birth [Table/Fig-4]. As pulmonary vascular resistance decreases, three scenarios can occur: insufficient collateralisation, leading to ischaemia and death; adequate collateralisation, causing "coronary steal" due to blood flowing retrogradely into the pulmonary artery from the right coronary artery; or massive collateralisation, which maintains sufficient perfusion even in the face of a steal phenomenon [3]. Due to the increased susceptibility of the single functioning coronary system to atherosclerosis, patients with ARCAPA may develop ischaemia later in life. Right-dominant coronary circulation is less tolerant of ARCAPA than left-dominant circulation.

The classic syndrome of infant myocardial ischaemia, infarction, and, if left untreated, death, is less common in cases of ARCAPA.



[Table/Fig-4]: Pathophysiology and direction of blood flow (Black arrow) seen in anomalous origin of the right coronary artery from the pulmonary artery [3]. (a) Soon after birth, high pulmonary pressure causes antegrade flow of deoxygenated blood through the right coronary artery. (b) Collateralisation between the left and right coronary arteries develops, causing dilation/tortuosity of both the right and left coronary arteries, as well as retrograde flow of blood through the right coronary artery into the pulmonary artery. (c) Immediately after aortic reimplantation of the right coronary artery onto the aorta, antegrade flow of oxygenated blood is established.

According to some authors, this is because the right ventricle has a lower oxygen demand than the left, and the RCA feeds a smaller portion of the myocardial territory than the LCA [3]. A comparison between ARCAPA and ALCAPA is shown in [Table/Fig-5] [3].

Variables	ARCAPA	ALCAPA
Incidence	0.002%	0.008%
Age of presentation	>2 years	<1 years
CHF	No	Yes
Ischaemia	No	Yes
Sudden death	Rare	Yes
PE	Murmur	CHF + /-SM
ECG findings	Non specific	Ischaemia Q Waves in 1 and aVL
Reimplantation	Yes	Yes

[Table/Fig-5]: Comparison between ARCAPA and ALCAPA.
SM: Systolic murmur; CHF: Congestive heart failure, ECG: Electrocardiogram, aVL: Augmented vector left

Soloff LA described four possible anomalous coronary artery connections to the pulmonary artery: ALCAPA, ARCAPA, origin of both coronary arteries from the pulmonary trunk, and origin of an accessory coronary artery from the pulmonary trunk [4]. The higher incidence of ALCAPA compared to ARCAPA is attributed to the proximity of the left coronary bud to the pulmonary artery sinus [4].

Imaging modalities for ARCAPA diagnosis include echocardiography, CCTA, MRI, and catheter angiography. Transthoracic Echocardiography (TTE) can delineate coronary ostia and assess flow. Intracoronary collaterals within the ventricular septum suggest ARCAPA on colour Doppler. Echocardiography can also demonstrate dilated coronary arteries and retrograde flow from the RCA to the pulmonary artery. ARCAPA can be mistaken for a coronary fistula if the RCA origin is not visualised. Transesophageal Echocardiogram (TEE) may be used when TTE is limited. CCTA and Cardiac MRI (CMR) provide detailed anatomical and functional information. Multiple collaterals from the Left Anterior Descending artery (LAD) and the Left Circumflex artery (LCX) to the RCA, which eventually drain into the pulmonary artery, can be better evaluated with coronary angiography. CMR can assess myocardial perfusion and fibrosis and measure the pulmonary-to-systemic blood flow ratio (Qp/Qs) [5].

When treating ARCAPA, there are two objectives: (1) to stop the condition's "coronary steal" and (2) to create a two-vessel coronary artery system that starts at the aorta [6]. To the authors knowledge, the present case represents a rare presentation of ARCAPA in South India, highlighting the role of multimodality imaging and successful surgical management.

CONCLUSION(S)

The ARCAPA is a rare congenital coronary anomaly that can lead to significant complications. In children with unexplained giddiness, correctly identifying the underlying pathology by systematically ruling out common causes, while maintaining a high index of suspicion for ARCAPA and performing timely imaging, can be life-saving.

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