RESEARCH PAPER

A rare case report: ALCAPA (Bland - White-Garland syndrome) - Congenital heart disease

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ABSTRACT
Background:Anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA), also known as Bland-White-Garland syndrome, is a rare but potentially fatal congenital heart defect. It constitutes approximately 0.25% to 0.5% of all congenital cardiac malformations, with a reported incidence of nearly 1 per 300,000 live births, particularly noted in the infant population (<1 year) in India.
Case Summary:
A 4 -month-old male child was presented with complaints of sudden onset increased work of breathing for the past one day, which worsened during feeding and was associated with increased irritability and sweating during feeds. There was no history of fever, cold, cough, or weight loss .Respiratory system examination was towards normal then referral to a pediatric cardiologist and subsequent echocardiography revealed a dilated dysfunctional left ventricle, severe mitral regurgitation (MR), moderate tricuspid regurgitation (TR), and a suspicion of Anomalous Left Coronary Artery from the Pulmonary Artery (ALCAPA) was confirmed by cardiac CT . The child was subsequently admitted and managed conservatively. Surgical intervention was later advised as part of the treatment plan.
Conclusion:The case underlines the importance of high clinical suspicion, early referral, and multidisciplinary care in managing congenital heart diseases like ALCAPA, where surgical correction remains the definitive treatment to prevent long-term cardiac complications and mortality.
Keywords: ALCAPA, Congenital Heart Diseases, Outcome
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INTRODUCTION

Anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA), also known as Bland-White-Garland syndrome, is a rare but potentially fatal congenital heart defect. It accounts for approximately 0.25% to 0.5% of all congenital cardiac anomalies, with an incidence of about 1 in 300,000 live births[1]. In this condition, the left coronary artery, which typically arises from the aorta, originates abnormally from the pulmonary artery, leading to myocardial ischemia, left ventricular dysfunction, and potentially life-threatening arrhythmias, as seen in a one-month -old child in India.

The pathophysiology of ALCAPA varies with age. In neonates, the pulmonary artery pressure is initially high and oxygenated, so perfusion may be adequate. However, as pulmonary vascular resistance decreases after birth, a "coronary steal" phenomenon develops, where oxygen-poor blood from the pulmonary artery is delivered to the left coronary system, causing myocardial hypoxia[2]. This can result in dilated cardiomyopathy, mitral regurgitation, or sudden cardiac death if left untreated[3].

Early diagnosis through echocardiography, cardiac MRI, or CT angiography is crucial. Surgical correction, typically by reimplantation of the left coronary artery into the aorta, has dramatically improved survival rates[4]. Here, we present the rare case of ALCAPA diagnosed in infancy, highlighting the clinical presentation, diagnostic challenges, and successful surgical management

Case Report: A 4-month-old male child was brought with increased work of breathing, irritability , inconsolable cry for 1day duration, There was history of suck rest cycle and sweating during feeding. No complaints of fever, cold, cough, weight loss. The child was initially taken to a nearby clinic, where nebulisation and symptomatic treatment were initiated.

On presentation, the child was in respiratory distress, tachycardic (HR: 175/min),tachypneic (RR: 74/min), and had oxygen saturation of 90% off oxygen. Systemic examination revealed muffled heart sounds, intercostal and subcostal retractions, and irritability. On Blood investigation, Hemoglobin level was normal, presence of mild leucocytosis, and other parameters were normal in

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range. On chest X-ray examination, no any abnormality found. 2D
echocardiography revealed a dilated dysfunctional left ventricle, severe mitral regurgitation
(MR), moderate tricuspid regurgitation (TR), and a suspicion of Anomalous Left Coronary Artery from the Pulmonary Artery (ALCAPA). LVEF had furtherdeclined to 15–20%. A CT coronary angiography validated the diagnosis, revealing anomalous origin of the left main coronary artery from the posterior pulmonary artery with normal contrast opacification, and no other major structural anomalies.
The patient was managed in Pediatric Intensive Care Unit (PICU) and started on
Heated Humidified High Flow Nasal Cannula (HHHFNC) oxygen support. Intravenous medications including milrinone, adrenaline, lasix, carnitine, thiamine, and aldactone were initiated, along with oral Vymada. Due to persistent respiratory distress, the patient was escalated to non-invasive ventilation (NIV) in pressure control mode and subsequently digitalized with digoxin. Levosimendan was administered over 48 hours based on cardiology input. Electrolyte imbalances were addressed, and cardiac medications adjusted as the child showed gradual clinical improvement after 3 days . Oxygen support was weaned, IV drugs transitioned to oral, then patient was improved and referred for surgical correction were initiated.

DISCUSSION:

This case of a 4-year-old male presented with acute onset respiratory distress, feeding intolerance, irritability, and diaphoresis highlights a rare and life- threatening congenital anomaly—ALCAPA presenting as acute decompensated heart failure in a previously asymptomatic 4 -month-old child. The timely identification through echocardiography and confirmation by CT angiography were crucial for diagnosis. Comprehensive PICU management with inotropes, diuretics, and respiratory support led to clinical stabilization.
In fetal life and early postnatal circulation, pulmonary artery pressure and oxygen content are sufficient to supply the myocardium. However, as pulmonary vascular resistance falls after birth, the perfusion pressure in the left coronary system decreases, resulting in retrograde flow of deoxygenated blood from the pulmonary artery to the left coronary artery. This "coronary steal" phenomenon causes chronic myocardial ischemia, progressive left ventricular dysfunction, and mitral regurgitation due to papillary muscle ischemia. [8] Our patient demonstrated all these features: dilated cardiomyopathy, severe MR, and reduced ejection fraction (LVEF 15–20%).
Diagnosis of ALCAPA can be challenging due to its variable presentation and nonspecific early symptoms. Echocardiography remains the first-line modality for diagnosis, but advanced imaging such as cardiac CT angiography or MRI provides definitive anatomical details. [9] In our patient, the diagnosis was confirmed through CT angiography, which showed anomalous origin of the left coronary artery from the posterior aspect of the pulmonary artery with preserved contrast opacification.
The mainstay of treatment is surgical correction, which typically involves reimplantation of the anomalous coronary artery into the aorta to restore a dual coronary system. [10] Medical management with inotropes, diuretics, and respiratory support serves as a bridge to surgery in decompensated cases. Our patient was stabilized in the PICU with high-flow oxygen, non-invasive ventilation, digoxin, milrinone, levosimendan, and diuretics was given and improved then patient was referred for definitive surgical intervention.
This case emphasizes the need for early recognition and high index of suspicion for ALCAPA in infants and children presenting with heart failure symptoms. Without
surgical correction, mortality approaches 90% within the first year of life, while successful surgical repair dramatically improves long-term outcomes and ventricular function. [11]

CONCLUSION:

The case underlines the importance of high clinical suspicion, early referral, and multidisciplinary care in managing congenital heart diseases like ALCAPA, where surgical correction remains the definitive treatment to prevent long-term cardiac complications and mortality.

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