

Repair of anomalous left circumflex dominant coronary artery originating from right pulmonary artery in an adult age patient: surgical correction with ligation and bypass graft: a case report

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ABSTRACT

Bland-White-Garland syndrome also known as anomalous origin of left coronary artery from pulmonary artery (ALCAPA) is a rare anomaly of coronary arteries among all congenital heart defects. Its clinical presentation at different age, either in children or adulthood has significant impact on outcomes leading to myocardial ischemia and heart failure. Early diagnosis of ALCAPA with the help of multislice CT angiography or detailed catheter study is always good for the patient to prevent the possible grave consequences. Surgical approach remains to be the definite treatment modality for ALCAPA with variable approaches and surgical methods to repair. We hereby present a case of 53 years male with ALCAPA from branched right pulmonary artery with collaterals between RCA and LCA.

Keywords: Anomalous coronary, ALCAPA, BWG syndrome

INTRODUCTION

Among all congenital heart diseases, anomalous coronary artery originating from pulmonary artery has been documented as one of the rare anomaly, which is being encountered in almost 1 in 300,000 cases worldwide.¹ This anomaly was first clinically described in 1933 by Edward Bland, Paul Dudley White, and Joseph Garland, also known as Bland White Garland syndrome or anomalous left coronary origin from pulmonary artery (ALCAPA).² This anomalous coronary origin leads to ischemia in left coronary system which further leads to ventricular dysfunction, mitral regurgitation and sudden cardiac death. This anomaly has been subdivided into two subtypes: infant and adult with its presentation at different age depending upon ischemia tolerance and collateral formation with age. Trans-thoracic imaging modality with other non invasive investigations CT angiography and magnetic resonance angiography can be used to identify the abnormality. Aim of the treatment for such anomaly remains confined to reconstruction for two separate coronary systems. There are several techniques for surgical repair including coronary button transfer, trans-pulmonary baffle creation or flaps, and coronary artery bypass grafting with ostial ligation. While some patients may still experience symptoms after the operation, the majority of patients recovers with their left ventricular function and is no longer at risk of sudden death. Further, severity of clinical presentation depends on steal of blood from coronaries and age.^{2,3}

We hereby present a rare case of anomalous partial left circumflex coronary originating from right pulmonary artery presenting in adulthood with mild to moderate heart failure features with successful surgical repair.

CASE

A male patient aged 53 years, presented to our hospital in cardiology unit with chief complaints of dyspnoea with abrupt onset of on and off pain in chest radiating to neck. On evaluation his blood pressure was recorded 132/84 mm of Hg, SpO₂ 98% at room air, heart rate of 92 beats per minute. On further examination, his trans thoracic echocardiography revealed mild RV dilation with LVEF 45-48% (Simpson's method). Chest X ray was normal and ECG was found to be in sinus rhythm. Considering his clinical presentation with insignificant findings, detailed cardiac evaluation was planned. CT Coronary angiography was performed, which revealed anomalous coronary with its ostia originating from right pulmonary artery (RPA) along with separate two normal coronary ostia originating from aorta as usual on left and right coronary positions along with co-dominance of left and right coronaries (**Figure 1**). Considering ambivalent results of CT coronary angiogram, further evaluation based on standard coronary angiography with cardiac catheterization study was performed, which revealed two separate normal coronary ostia for right and left coronary arising from aorta

along with anomalous coronary connection between RPA and left circumflex coronary artery (LCX). It also revealed co-dominance of both left and right coronary system with significant collaterals connecting both sides (**Figure 2**). As per cardiac catheterization results, patient was planned for surgical intervention in view of persistent symptoms.

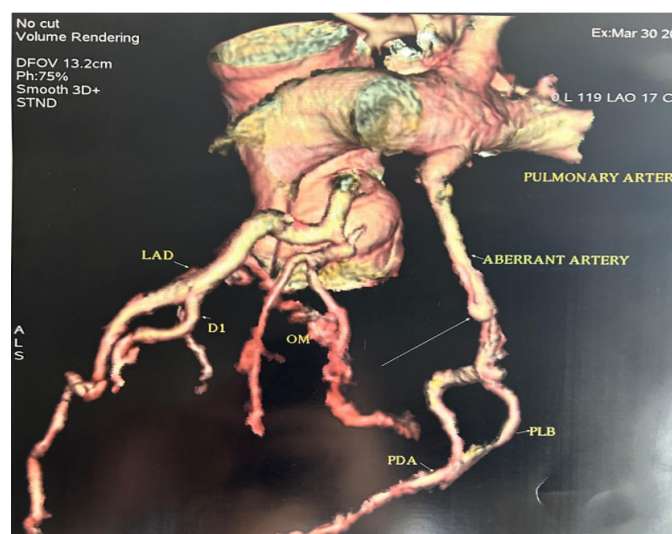


Figure 1. CT images of coronary angiogram showing anomalous coronary origin from right pulmonary artery
CT: Computed tomography, PDA: Patent ductus arteriosus



Figure 2. Conventional images of coronary angiogram one catheter placed in main pulmonary artery from right atrium, while other catheter spilling contrast into left main ostia with its spillage further into right pulmonary artery through a separate origin of dominant left circumflex coronary artery co-dominant system with collaterals between left and right coronaries

Surgical Procedure

Standard midline sternotomy was done. Aorto-bicaval cannulation was done. Cardiopulmonary bypass (CPB) was established with mild hypothermia of 32 degrees. RPA was dissected and anomalous connection was found between RPA and left circumflex coronary origin running behind aorta. Initial plan was to re-implant coronary ostia to aorta directly, considering the length of anomalous coronary was found short and tortuous course of coronary running behind aorta into posterior myocardium, plan of surgical repair from coronary re-implantation was converted to ostial ligation and bypass graft (**Figure 3**). Coronary ostial end was identified, looped around and snuggled for 5 minutes

and checked for any ECG changes. This depicted mild ECG changes with ST elevation of 0.06 mm in antero-lateral leads. Considering mild ECG changes, coronary artery bypass with one saphenous venous grafting was done for obtuse marginal vessel with proximal end anastomosed to aorta. Anomalous left circumflex coronary ostial connection originating from RPA was ligated juxta-pulmonary origin externally and clipped. CPB was weaned on minimal inotropic support. Total CPB time of 58 minutes with cross clamp time was 25 minutes. The postoperative course was uneventful, and patient was extubated on the first postoperative day. He was discharged on the fifth postoperative day. Patient is doing well with his 10 months follow up without any symptoms and preserved ejection fraction.

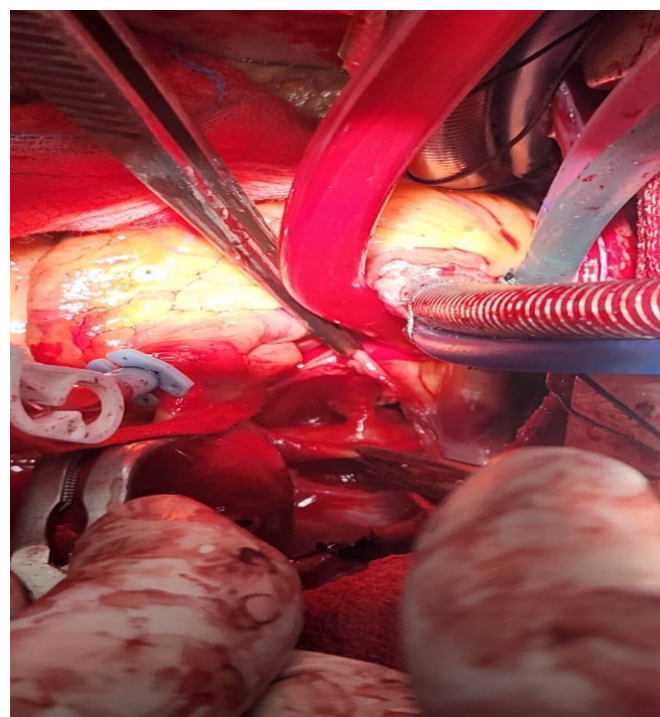


Figure 3. Intra-operative image depicting anomalous coronary ostia of dominant left circumflex originating from right pulmonary artery as shown in image between forceps holding tissue

DISCUSSION

Based on presentation of age, ALCAPA patients are typically classified into two types depending on the presence of collaterals:

- The adult type for those with well-established collaterals and
- The infantile type for those without collaterals.

Certain factors such as the timing of the closure of the patent ductus arteriosus (PDA), the pressure gradient between the pulmonary and coronary arteries, and the development of collaterals, as well as the type of coronary anatomy plays a vital role in presenting symptoms of the patient.^{3,4} Surgery is recommended in patients with ALCAPA, even in the absence of symptoms or a significant left-to-right shunt syndrome, given the risk of ventricular arrhythmias and sudden death.³ Intracoronary collateral arteries benefit such patient for survival beyond infancy.⁵ Multislice CT angiography plays pivotal role for non-invasive imaging with its spatial resolution for diagnosis of abnormal coronary arteries and their course.^{4,5} Aim of surgical treatment for ALCAPA

patient remains confined to establish a two coronary vascular system to provide oxygenated blood to entire heart. Various surgical methods depending upon coronary origin from pulmonary trunk has been performed over the time and such surgical techniques are used depending upon variable anatomy, which includes direct re-implantation of coronary to aorta (coronary button transfer), intra-pulmonary baffle repair (the Takeuchi technique), subclavian coronary artery anastomosis and coronary artery bypass grafting with ostial ligation. Heart transplant remains reserved for the patient presenting late with poor ejection fraction.

It has been found in past by treating such patients may still experience symptoms after the operation, despite majority of patients recover left ventricular function and are no longer at risk of sudden death.^{6,7} In a case series reported by Mishra et al.,⁸ the outcomes of 105 patients with anomalous origin of the coronary artery from the pulmonary artery and 98 patients with ALCAPA; the median age at operation was 5.8 months with a median follow-up of 5.9 years. All patients underwent coronary re-implantation. In-hospital mortality was reported to be 8.5%, with no reported late deaths. In the Mishra series, 33.6% of patients who underwent surgery during the newborn period (<6 months of age) had good recovery of left ventricular function (mean left ventricular ejection fraction from 50 to 55%) and had improvement in mitral regurgitation from moderate severity to mild residual regurgitation. In addition, 52% of the patients who underwent operations late in infancy (after 6 months of age) had residual impairment of left ventricular function (mean left ventricular ejection fraction from 40 to 50%) and mild-to-moderate mitral regurgitation.⁸

Another review study of 98 infant ALCAPA cases, Radman et al.⁹ found that approximately three years after corrective surgery, left ventricular function returned to normal in 98% of cases, whereas the trajectory of mitral regurgitation was more difficult to predict. The uncertainty regarding the improvement of mitral regurgitation was thought to be caused by potentially irreversible preoperative ischemia or suboptimal coronary perfusion after surgical correction due to loss of patency in the left coronary artery.⁹

The type of surgical correction being done, invariably impact the outcome and postoperative complications.

Surgical ligation when performed can lead to re-canalization and persistent silent ischemia with mitral regurgitation and may sometimes lead to sudden death.

Direct re-implantation technique when performed in infancy has good outcomes, but at the same when performed in adult age can lead to coronary artery tearing and bleeding, as it has decreased elasticity and high chance of kinking ostia during its mobilization.

Pulmonary baffle technique (Takeuchi repair) can lead to supra-valvular pulmonary artery obstruction, coronary-pulmonary fistula or aortic regurgitation.

Coronary artery bypass grafting in adult has been found to present promising results with internal mammary artery and saphenous vein graft with good calibre.^{8,10}

Very few patients may present with residual symptoms due to irreversible myocardial damage and ischemia prior to surgery. At the same, a small subset of patients with poor ejection fraction may need implantable cardioverter defibrillators (ICD) in order to support left ventricle and dysarrhythmias in post operative period. Regular follow up is required in postoperative duration with monitored cardiac function, including ECG, echocardiogram and cardiac magnetic resonance imaging which help in long term outcomes.

Further survival of a patient depends on presenting illness along with factors such as age of presentation, lung maturity, the ratio of the left and right shunting, the development of collateral circulation, and the predominance of the right coronary artery and opting appropriate surgical method at correct time. Surgical treatment was chosen for our patient based on age, good collateral development and based on symptoms, ostial ligation with coronary graft was applied following this patient had uneventful outcome with early recovery.^{7,11}

CONCLUSION

We hereby conclude that considering every individual as separate entity with such cardiac anomaly, one should plan for patient looking at his diagnostic approach followed by appropriate method of surgery for survival benefit along with best outcomes for ALCAPA patients. Since over the past such anomaly has been found with anomalous origin of coronary arising from main pulmonary trunk but in our case it has been found arising from right branch of pulmonary trunk, which can be helpful for surgeons treating such condition in future. Such need of treatment plan helps not only patient but surgeon as well depending upon his best outcomes.

ETHICAL DECLARATIONS

Informed Consent

Written informed consent was obtained from the patient included in this report. Signed consent forms are retained by the authors and are available upon request.

Peer Review Process

This report underwent external peer review.

Conflict of Interest

The authors declare no conflicts of interest.

Financial Disclosure

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Author Contributions

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