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Anomalous Left Anterior Descending Coronary Artery Arising from the Pulmonary Artery (ALADAPA): A Narrative Review and the 52nd Case in the World

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Abstract

Background: Congenital coronary artery abnormalities are detected in 0.6 to 1% of adult patients having coronary angiography. Anomalous origin of coronary arteries from the pulmonary artery is a rare congenital anomaly that is almost always lethal if not diagnosed and treated. Only 51 cases of ALADAPA have been reported in the literature, with clinical presentations varying from sudden cardiac arrest to incidental murmur depending on the area of the myocardium affected.

Case Presentation: A 32-year-old female who presented with unstable angina was diagnosed with an anomalous origin of the left anterior descending artery from the pulmonary artery (ALADAPA). The patient underwent uneventful coronary artery bypass grafting where the left internal mammary artery (LIMA) was anastomosed to the left anterior descending coronary artery (LAD) and the abnormal origin of ALDAPA was ligated. The patient's symptoms improved remarkably. Computerized tomography coronary angiography performed six weeks postoperatively showed patent LIMA to LAD.

Conclusion: LIMA to ALDAPA can be performed successfully with excellent short-term results. Reporting these rare conditions to cardiologists and cardiac surgeons will provide further insights into their management.

Key Words: Coronary Artery Anomalies, Anomalous Left Anterior Descending Artery from Pulmonary Artery (ALADAPA), Anomalous Left Coronary Artery from Pulmonary Artery (ALCAPA), Left Internal Mammary Artery (LIMA), Coronary Artery Bypass Grafting (CABG).

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Background

Normal coronary artery distribution includes the right coronary artery (RCA) arising from the right aortic sinus of Valsalva and the left coronary artery (LCA) arising from the left aortic sinus of Valsalva.[1]

Congenital coronary artery abnormalities are detected in 0.6 to 1% of adult patients undergoing coronary angiography. [2] Anomalous origin of coronary arteries from the pulmonary artery is a rare congenital anomaly that is almost always lethal if not diagnosed and treated. Of these anomalous left main coronary artery from pulmonary artery (ALCAPA) is the most common anomaly with an incidence of 1 in 30,000 to 1 in 300,000 people. Other variants are extremely rare including anomalous LAD (ALADAPA), circumflex and RCA.[3,4]

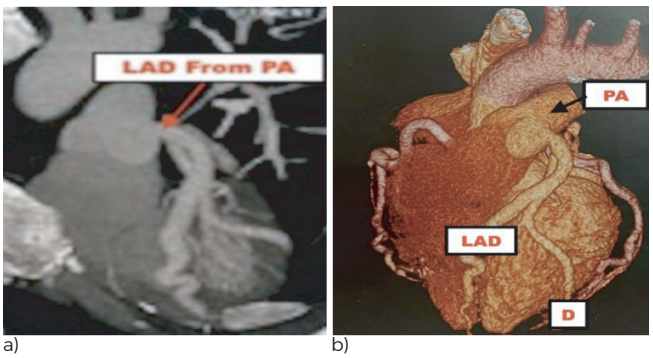
Only 51 cases of ALADAPA have been reported in the literature, with clinical presentations varying from sudden cardiac arrest to an incidental murmur depending on the myocardial territory affected.[4]

Case Presentation

This is a 32-year-old female patient who complained of typical chest pain at rest (unstable angina, Canadian cardiovascular society class IV). No previous history of chronic illnesses and no known family history of coronary or congenital cardiac diseases. Physical examination was unremarkable except for a soft systolic murmur over the left parasternal area. Her laboratory investigations were within the normal range. Resting electrocardiogram (ECG) did not show any specific changes, and exercise ECG was positive for ischemia. Echocardiography showed preserved left ventricular function (LVF) with no valve abnormalities.

Percutaneous coronary angiography showed a large dominant, ectatic and tortuous RCA vessel, with numerous large collaterals to the LAD shunting to the PA. LMCA origin was normal and bifurcating into left circumflex (L.Cx) and small proximal LAD stump. The LAD was also dilated and abnormally originating from the main PA trunk.

Computerized tomography coronary angiography (CTCA) confirmed the diagnosis of anomalous LAD arising from the PA with normal origins of RCA and L.Cx. (Figure 1 a,b). The case was discussed by the cardiac team



a) b)

Figure 1 (a,b): 3-D reconstruction of a coronary CT angiogram showing the LAD arising directly from the main pulmonary artery rather than the left coronary sinus.

and a decision was taken to perform LIMA-to-LAD and disconnect the abnormal origin of LAD from the PA.

Through median sternotomy, the left internal mammary artery was harvested, followed by full heparinization and commencement of cardiopulmonary bypass. The operative findings confirmed large ectatic LAD (3-4 mm) arising from the left lateral aspect of the main pulmonary artery trunk with palpable thrill over the PA (Figure 2). The RCA was large and tortuous giving numerous large collaterals to LAD.

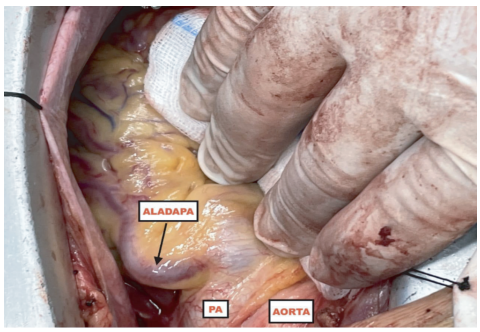


Figure 2: Abnormal LAD arising from pulmonary artery (PA).

Trial of off-pump coronary artery bypass grafting (CABG) was aborted due to torrential collateral flow which rendered visibility extremely difficult and necessitated conversion to on-pump, beating heart LIMA-to-LAD anastomis, that was performed using 7-0 Prolene (Figure 3).

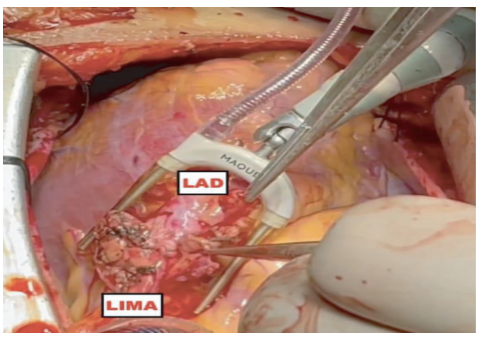
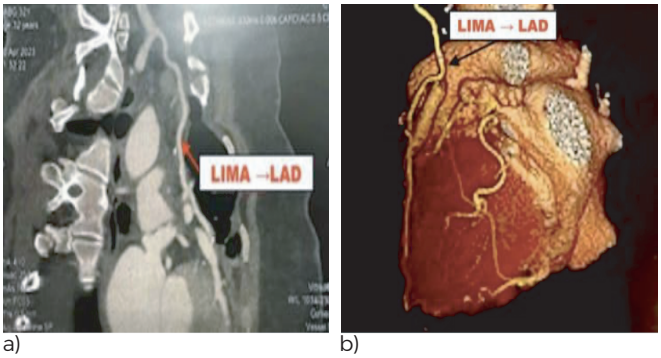


Figure 3: LIMA to ALADAPA done on pump beating heart.

The origin of LAD from PA was ligated using double armed 3-0 proline stitches. Weaning from bypass was very smooth and standard closure was completed. Postoperative recovery was uneventful and the patient was discharged on the fourth postoperative day.

One month later, on the 1st postoperative visit, the patient reported marked improvement of symptoms with no angina at all. CTCA was done 6 weeks after surgery and showed patent LIMA to LAD with good distal run off and no shunt to PA (Figure 4 a and b).



a) b)

Figure 4: **a, b** Shows patent LIMA to LAD 6 weeks post-operatively.

Discussion

Anomalous coronary artery arising from the pulmonary artery is a rare but serious congenital cardiac anomaly. Without surgery, approximately 85% of patients die before reaching adulthood.[5]

In a systematic review conducted in 2019, 51 cases of ALADAPA were described with mean age 27.3 years with female predominance. Clinical presentation was variable ranging from angina, as in this patient, to palpitations and heart failure. Fatigue and fever were reported as incidental findings in a few cases (approximately 10%) and sudden cardiac death was reported in three cases.[4] The development of symptoms depends on the degree of collateralization. Initial collateralization is essential for survival to adulthood.

However, as collaterals become larger with less resistance, they create a steal syndrome from the area of supply and shunt significantly to the pulmonary artery. Conventionally, ALCAPA and similar anomalies have been diagnosed by autopsy or contrast echocardiography. Both invasive coronary angiography and non-invasive coronary CTA are considered equivalent and are the main diagnostic methods in today's practice. Magnetic resonance imaging can provide the same data as CT with the added benefit of myocardial perfusion.[6] Additionally, short LAD originating from the aorta terminating early in the interventricular groove creating "Dual LAD "pattern have been reported by CT studies.[7]

Surgical correction should be considered even in asymptomatic cases given the risk of sudden death. Re-implantation of LAD button onto the aorta may be considered as a treatment option.

However, LIMA to LAD with proximal ligation of the anomalous artery is an accepted alternative whenever transfer is not feasible in certain cases such as deeply buried proximal LAD or originating from the far lateral side of the PA.[8]

Left internal mammary artery (LIMA) is traditionally preferred although no long-term data comparing it to the vein are available in this category of patients. In one case report the anomalous LAD was using a saphenous vein graft with a follow up angiography performed 3 years later, revealing a patent venous graft and regression of collaterals.[9] Simple ligation of the anomalous vessel will abolish the shunt but will leave extensive collaterals intact, creating a dead space in the coronary circulation with a risk of future occlusion and ischemia.

Other reported techniques include intrapulmonary tunnel repair (Takeuchi repair) which carries the risk of tunnel occlusion and supravalvular pulmonary stenosis.[10] Furthermore, the use of 8 mm Dacron tube graft interposed between LAD and aorta due to failed mobilization of LAD button was reported. The patient was asymptomatic postoperatively, but unfortunately the graft showed stenosis on coronary CT-angiography on follow up.[11]

Conclusion

This is a report of the 52nd case of ALADAPA worldwide treated by direct LIMA to LAD with excellent short-term results. Reporting successful surgery for these rare cases to cardiologists and cardiac surgeons provides further insights into their management.

Disclosure Statements:

The authors have no conflicts of interests to declare.

Ethics Approval and Consent to Publish:

The Ethics and Research committee, Medical directory, Alshefa Hospital, approved the case report study (committee's reference number CR1029/2023)

Consent for Publication:

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in- Chief of this journal.

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