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ALCAPA Associated with Congenital Lobar Emphysema: A Very Rare Association

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Abstract

We describe a rare association of anomalous left coronary artery from pulmonary artery (ALCAPA) with Congenital lobar emphysema (CLE) in a five month old male infant. The patient presented to our hospital with lower respiratory tract infection. Incidental detection of cardiomegaly and hyperinflation on chest X-ray was further evaluated with echocardiography and CT scan. Child was given a final diagnosis of ALCAPA with CLE right upper and middle lobe. He underwent corrective surgery for both the condition.

Introduction

This report highlights a rare association of ALCAPA with CLE of right upper and middle lobe in a five month male infant. This patient presented with features of lower respiratory tract infection. Incidental detection of cardiomegaly and hyperinflation of right lung on chest x-ray lead to the diagnosis of ALCAPA with CLE. The patient underwent surgical correction for both the conditions, requiring prolonged venovenous ECMO (Extra corporeal membrane oxygenation) for respiratory failure.

Case Report

Five month male infant was born to a second gravida through normal vaginal delivery, having a birth weight of 3 kg. He had an uneventful perinatal period. He presented with a history of fast breathing noticed since one month of age, high-grade intermittent fever, respiratory distress and decreased feeding for the past one week. Clinically he was tachypneic and febrile with a heart rate of 146 beats per minute, blood pressure of 68/45 mmHg. Chest examination revealed bilateral crackles with scattered wheeze. Cardiovascular examination revealed normal S1 and S2, audible S3 present and no murmurs.

Routine blood investigations showed raised total leucocyte counts with neutrophilic predominance), biochemical parameters were within normal limits. Procalcitonin was 10 ng/ml. Electrocardiogram (Figure 1) showed prominent deep q waves in lead I, avL and V5. Chest X-ray showed bilateral non homogenous opacities, hyperinflation (prominent over right upper and middle zone) and cardiomegaly. Echocardiogram done showed severe left ventricle (LV) dysfunction with global hypokinesia and ejection fraction of 15%. Moderate mitral regurgitation was present with hyperechogenicity of endocardium and papillary muscles. Right coronary artery was dilated and tortuous while left coronary artery was not seen. There were multiple intercoronary collaterals present. Contrast enhanced CT scan done showed right upper and middle lobe lobar emphysema with bilateral basal atelectasis. Right coronary artery was dilated and left coronary artery was seen arising from the non-facing sinus of main pulmonary artery (Figures 2 and 3).

Patient was started on broad spectrum antibiotics and high flow nasal oxygen therapy. He was ventilated electively for worsening respiratory distress. He was given seven days of broad spectrum antibiotics, ventilatory and ionotropic support (dobutamine) with no improvement. After one week of antibiotics patient was taken up for corrective surgery. On cardiopulmonary bypass using aortobicaval cannulation, right upper and middle lobectomy was performed (Figure 4). The left coronary artery was arising from the non-facing sinus of pulmonary artery and was intramyocardial immediately after takeoff. An autologous untanned pericardial tube of 4 mm diameter was constructed to elongate the coronary artery and implanted in the left coronary sinus of aorta. The Pulmonary artery was reconstructed using autologous pericardium (Figure 5). The patient was successfully weaned off bypass and was hemodynamically stable on moderate inotropic support. Over the next two post operative days there was consolidation of the left upper lobe. The ventilator parameters became prohibitive to adequate cardiac output and HFOV (High frequency oscillatory ventilation) was attempted, which was not tolerated well. He was placed on elective venovenous ECMO on the third post operative day. The patient did not show any improvement and the lung did not show any recovery. After seven days of venovenous ECMO the baby was deemed as irreversible lung injury and ECMO support was withdrawn in consultation with the parents to a fatal outcome.

Discussion

ALCAPA generally is an isolated anomaly, but may also be associated with patent ductus arteriosus, ventricular septal defect, tetralogy of Fallot and coarctation of the aorta [1]. ALCAPA association with CLE is extremely rare with only one published case report [2]. Our case was picked up based upon chest X-ray finding of cardiomegaly and hyperinflation. The diagnosis was further confirmed on echocardiography and contrast enhanced CT.

Our patient came in severe sepsis, which did not resolve completely even after a week of appropriate antibiotics. We had to take up the patient for surgery because of rapid deterioration on supportive management. He had a negative outcome post operatively due to respiratory failure inspite of an attempt of VV ECMO support.

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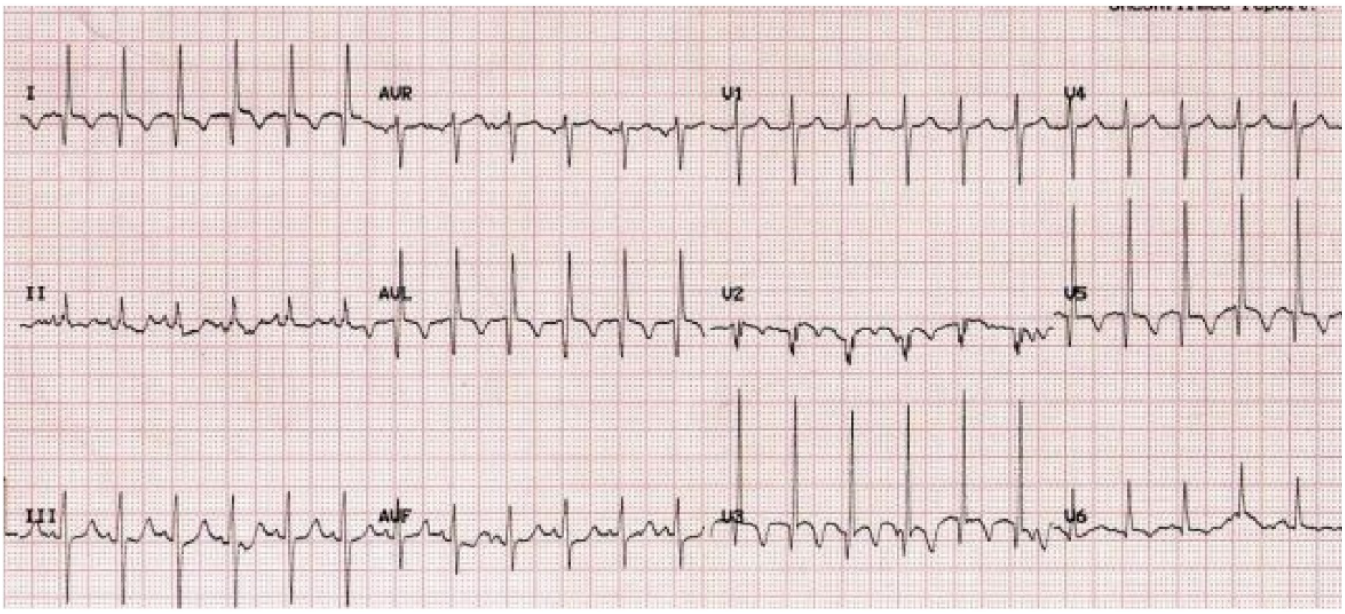

Figure 1: ECG showing deep Q waves in leads I, AVL and V5.

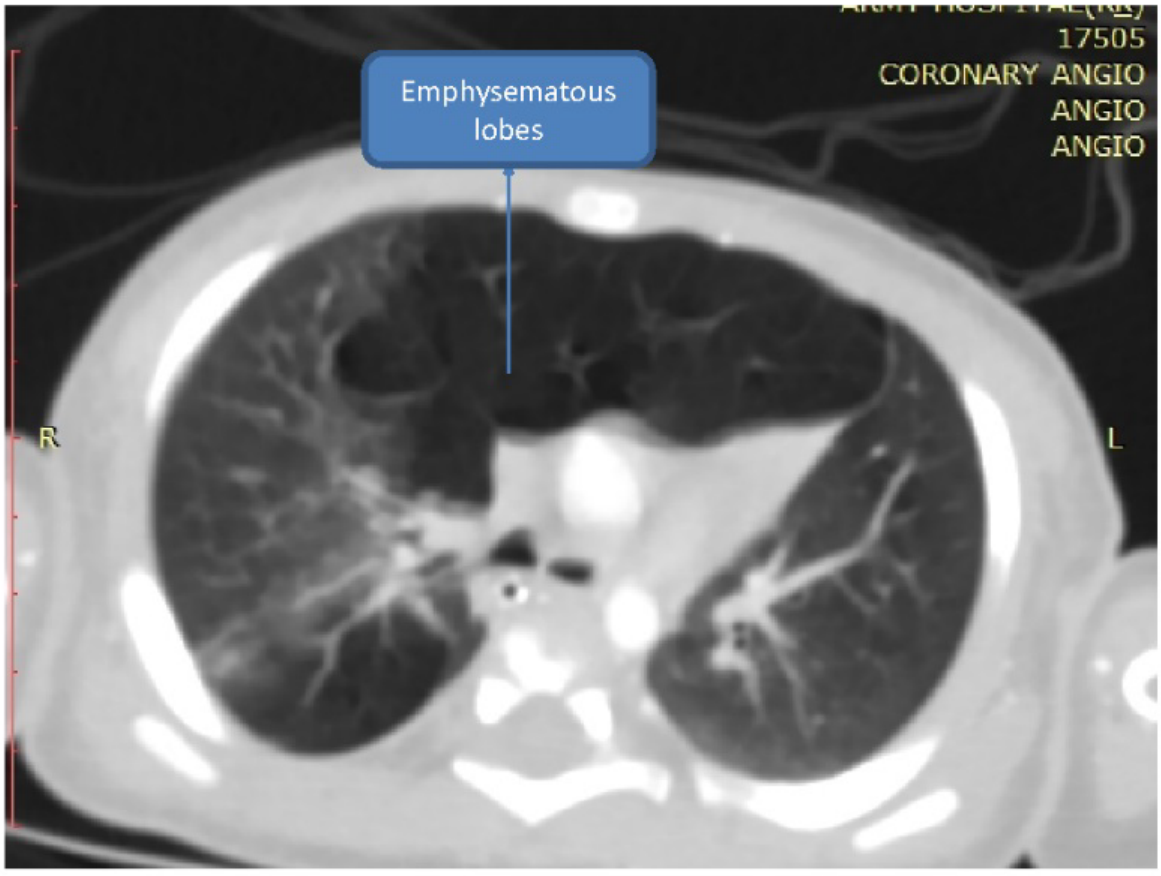

Figure 2: Contrast enhanced CT scan axial section showing emphysematous lobes in right lung.

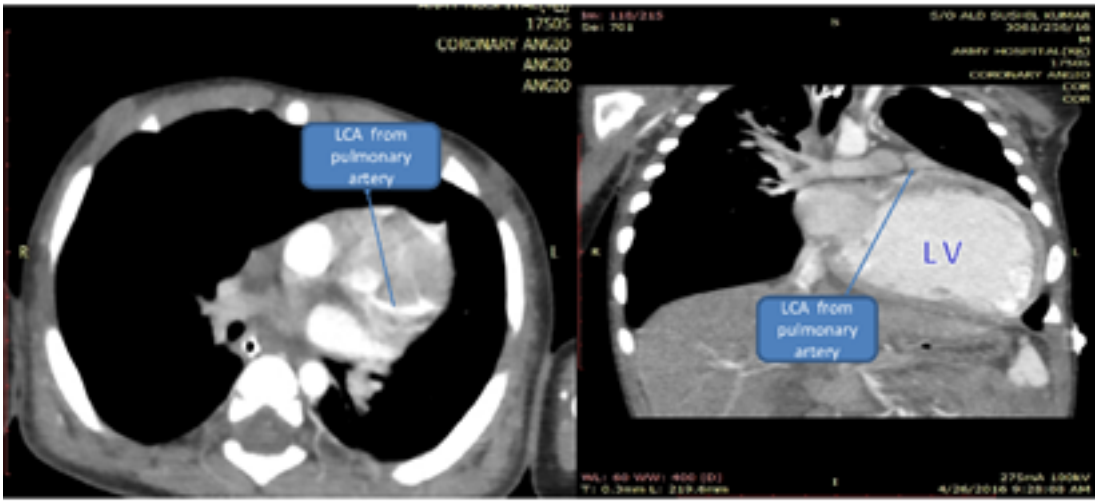

Figure 3: Axial and coronal section of CT scan showing origin of left coronary artery (LCA) from pulmonary artery.

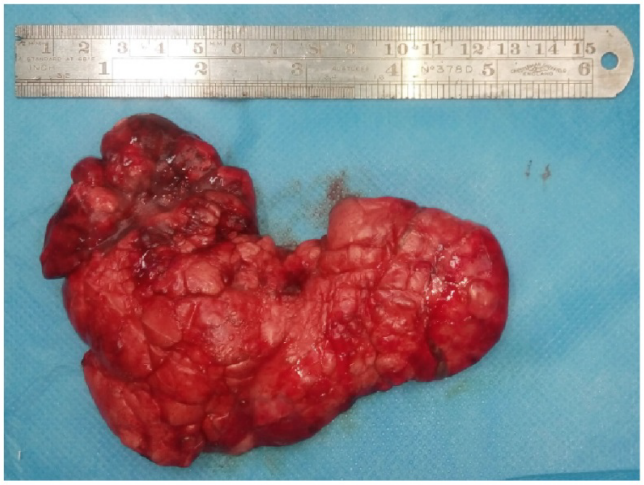

Figure 4: Restricted right upper and middle lobes.

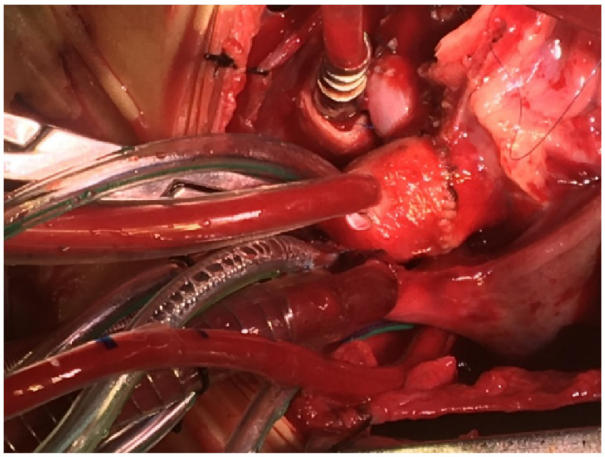

Figure 5: Showing reimplanted left coronary artery on to the aortic sinus.

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