A Young Infant with Large Anomalous Systemic Artery to Supply Left Lower Lobe of the Lung with Normal Intracardiac Anatomy

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Abstract

Background: Isolated **a**nomalous systemic arterial supply to the lobe of the lung in patients with normal intracardiac anatomy is a rare congenital pulmonary vascular anomaly. The diagnosis can be easily overlooked or delayed. The treatment can be arterial embolization, simple ligation, anatomical segmentectomy, lobectomy. If left untreated it can cause hemoptysis, pulmonary hypertension, and heart failure in adulthood.

Case presentation: We present a young infant with isolated large anomalous systemic arterial artery to left lower lobe of the lung. The initial diagnosis was raised by echocardiogram at 4 months of age for a murmur evaluation. It was confirmed by CTA. The patient has persistent left side heat volume overload. At 14-month of age the patient had Device occlusion of this large artery with two 10 mm Amplatzer vascular plug II devices.

Conclusions:

Anomalous systemic arterial supply to the lobe of the lung is a rare congenital condition. It can be detected at early age by echocardiography and more advanced image modality. Device occlusion of this vessel can be successfully achieved in young infant.

Keyword: Anomalous systemic arterial supply, device occlusion, embolization

Introduction

Anomalous systemic arterial supply to the lung lobes is a rare congenital pulmonary vascular anomaly. There are only handful cases reported, most in adult patients. (1,2, 3)

Case Presentation

The patient was born at 31 weeks of gestation with intrauterine growth restriction. At 4-month of age he presented for a cardiology evaluation for a continuous murmur evaluation. An echocardiography reveals a mild left side heart dilatation with mild mitral valve insufficiency, otherwise normal intracardiac anatomy. (Figure 1)

However, a large vessel was noticed originating from the descending aorta at level of low left atrium coursing towards left lower lobe of the lung. There is abundant left lower pulmonary venous return noticed. Further CTA of the chest confirmed this large anomalous artery measuring 6 mm. (Figure 2) There is no sequestration on CTA. The patient has persistent left side heart volume overload on serial echocardiographs and is mildly symptomatic. At 14-month of age the patient was referred for catheterization and possible embolization. The angiography reveals the central left pulmonary artery supplies the left upper lobe which drains normally to the left upper pulmonary vein and the large, anomalous vessel supplies the entire left lower lobe. Therefore, successful device occlusion was attained with two 10mm Amplatzer vascular plugs. (Figure 3) The patient has been doing well since then.

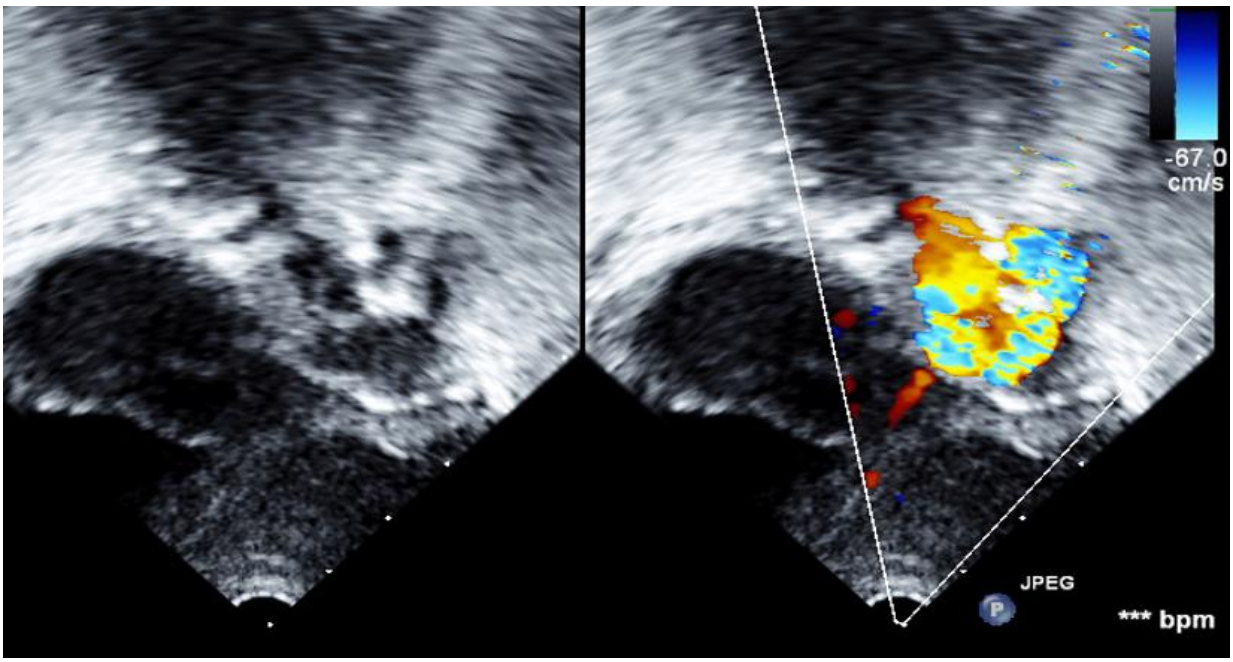

Figure 1 A large vessel seen from descending aorta coursing toward left lower lobe of the lung behind left atrium from subcostal view.

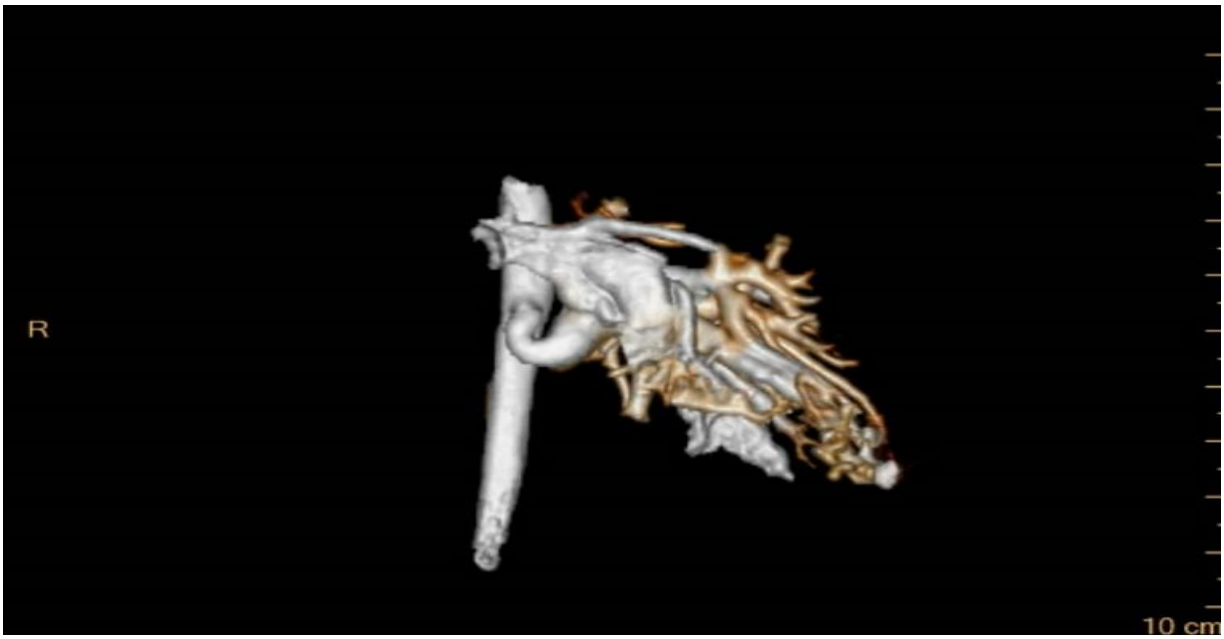

Figure 2 The large anomalous systemic artery from descending aorta coursing towards the left low lobe of the lung on 3D reconstruction of CTA

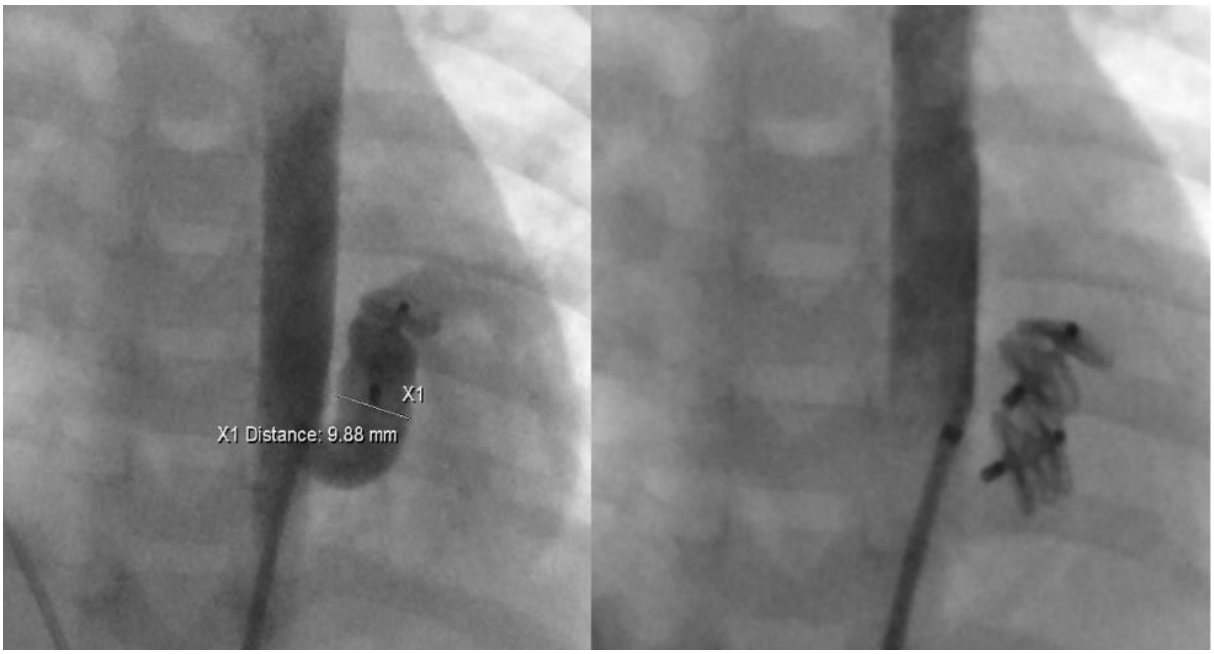

Figure 3 Angiography before and after the Device occlusion of this large artery with two 10 mm Amplatzer vascular plug II devices.

Discussion

An anomalous systemic arterial supply to the lobes of the lung is a rare congenital condition with basal lobe being the most common involved. (4,5) Many terms have been used to describe this condition since there is lack of consensus. The most widely accepted term is “anomalous systemic arterial supply. (6, 7) There are very limited case reports, most in adults. One similar case report was reported by Do Wan Kim et al in a 7-month-old infant who received end-to-side anastomosis between the abnormal systemic artery and left pulmonary artery. (8) The diagnosis of this condition can be easily missed or delayed. No standard treatment has been established. (9) Initially , the main reported treatment was surgical resection of the involved lung after ligation of the anomalous artery; however the trend shifted towards trans-arterial embolization. (10)

Conflicts of Interest

The authors declare that they have no conflicts of interest.

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