

## Anomalous Systemic Arterial Supply to Normal Basal Segments of the Left Lower Lobe of the Lung: Therapeutic Implication

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### ABSTRACT

We describe a 5-month old infant who presented with a continuous murmur and enlargement of the left heart. The patient's diagnosis was an anomalous systemic arterial supply to basal segments of the left lower lobe characterized by a lack of a pulmonary arterial supply. This condition was treated without lobectomy. To our knowledge, this report is the first to describe an anomalous systemic arterial supply to basal segments of the lower lobe of the left lung with a single arterial supply that was treated in childhood without lung resection. Our case offers an alternative treatment to surgical lobectomy for this abnormality.

### INTRODUCTION

An anomalous systemic arterial supply to the basal segments of the left lung without sequestration in which the normal pulmonary arterial supply is absent is a rare congenital anomaly. This condition is usually asymptomatic in children but may be discovered following an incidental finding of a continuous chest murmur [Ishihara 1979] and left ventricular failure that has developed from a left-to-left shunt [Yabek 1981]. Patients with this anomaly usually have been treated with lobectomy [Ishihara 1979; Yabek 1981]. We describe the case of an infant who presented with left heart failure that was caused by an abnormal systemic arterial supply as a single source to the left lower lobe of the lung that arose from the descending aorta. This anomaly was treated without lobectomy.

### CASE REPORT

A 5-month-old girl was referred to our institution because of a heart murmur. During the clinical examination, a continuous murmur was detected at the left precordium and was more prominent at the back of patient. The chest radiograph

revealed mild cardiomegaly. An echocardiographic evaluation revealed left atrial and ventricular dilatation. The systolic functions of the left ventricle were normal. Color Doppler imaging revealed an increased pulmonary venous blood flow and an aberrant artery arising from the descending aorta that displayed continuous flow at the cardiac level (Figure 1). Multidetector computed tomography (CT) images revealed a normal bronchial tree and a tortuous artery that arose from the descending aorta and supplied the basal segments of the left lower lobe of the lung (Figure 2). Although we carefully evaluated the selective left pulmonary arteriogram, we found no normal pulmonary artery that supplied the basal segments of the left lung (Figure 3A). An aortographic examination confirmed the presence of this tortuous and dilated vessel that arose from descending aorta and supplied the basal segments of the left lung. This vessel drained into the left atrium (Figure 3B). A left posterolateral thoracotomy was performed, and the aberrant artery was clamped. After clamping, we observed no change in the color of the basal segments over a 45-minute period. Because we noticed no remarkable changes in the color or shape of the affected segments of the lung, we canceled the preoperative plan to resect the affected segments of the lung. The aberrant artery was simply ligated and divided.

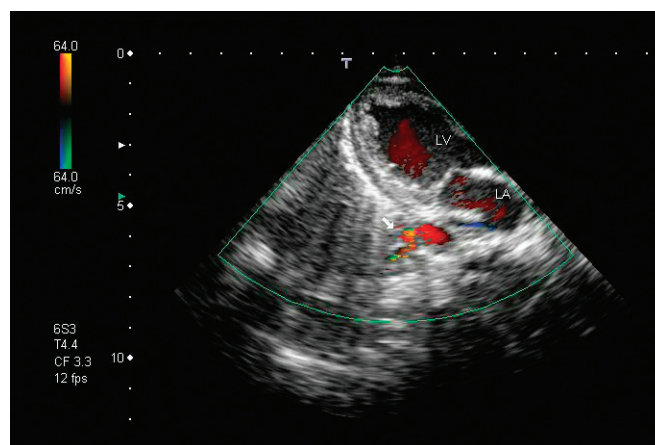


Figure 1. Color Doppler imaging shows increased pulmonary vein blood flow and an aberrant artery arising from the descending aorta and showing continuous flow at the cardiac level.

Received January 17, 2008; accepted March 17, 2008.

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Figure 2. Multidetector computed tomography image showing a tortuous artery arising from the descending aorta.

The patient had an uneventful recovery and was discharged on the sixth postoperative day. Routine clinical, chest radiography, and echocardiography examinations were performed at 1, 3, 6, 12, and 16 months after the surgery. A selective left pulmonary arteriographic evaluation performed 6 months

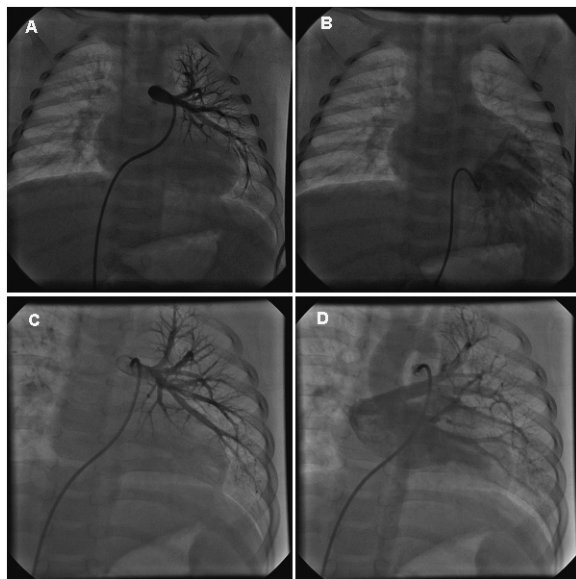


Figure 3. Initial angiograms. A, Selective left pulmonary angiogram showing no vessels to the left lower lobe. B, Thoracic aortogram demonstrating that the anomalous systemic artery arising from the descending thoracic aorta supplies the affected segments. C, A selective left pulmonary angiogram obtained 6 months after the ligation procedure shows the lack of vessels in the left lower lobe. D, A late phase of the selective left pulmonary angiogram reveals venous drainage and the whole systemic circulation with no vessel to the left lower lobe of the lung.

after the procedure revealed no arterial supply to the left lower lobe from the pulmonary and systemic arteries (Figures 3C and 3D). A pulmonary perfusion scintigraphy examination performed 16 months after the surgical procedure to evaluate the circulation of the left lower lobe showed the perfusion defect. A pulmonary CT scan performed at the same admission revealed no effusion, atelectasis, or mass (Figure 4). The patient has been followed uneventfully for 18 months.

## DISCUSSION

The systemic arterial supply to the basal segment of the lower lobe without intralobar sequestration has been categorized as type 1 sequestration according to Pryce's nomenclature [Pryce 1946]. The aberrant systemic artery either may provide a partial arterial supply in addition to the normal pulmonary arterial supply or may be the only arterial supply to the lung, as in our case. Even with the advent of new imaging modalities, diagnosis of this anomaly in early childhood period is very rare, and patients with this anomaly usually receive their diagnoses as adults, when they present with hemoptysis and recurrent pulmonary infections [Yamanaka 1999; Baek 2006]. This anomaly should be suspected in pediatric patients who present with a continuous murmur or in the absence of known causes of left heart dilatation. The usual choice of treatment is division of the anomalous artery followed by resection of the involved lung lobe [Ishihara 1979; Yamanaka 1999]. Recently, Baek et al [2006] described an adolescent patient who had been treated with simple ligation without lung resection. These investigators noted that the presence of the normal pulmonary artery in the diseased segments of the lung was hard to define with pulmonary angiography, largely because of the flow from the anomalous artery. Pulmonary arteriography is accepted as the gold standard



Figure 4. Computed tomography scan of the left chest 16 months after the surgical procedure showing a normal left lung.

for demonstrating the presence of a normal pulmonary arterial supply in affected segments, and we did not observe a pulmonary artery supplying the left lower lobe, either before or after ligation of the aberrant artery. The occlusion of an aberrant artery arising from the aorta in a dually supplied lung has been successfully performed in catheterization laboratories [Brühlmann 1998]. In our case, we ligated the aberrant artery without lung resection, even though the pulmonary arterial supply was lacking. Our patient has been free of symptoms, and the results of chest radiography, echocardiography, and lung CT examinations were within normal ranges at the last follow-up 16 months after surgical ligation. This outcome could be attributed to the bronchial blood supply alone being sufficient to provide adequate oxygen and nutrition to the tissue of the left lower lobe. To our knowledge, this report is the first to describe a single anomalous systemic arterial supply to basal segments of the lower lobe with a single arterial supply that was treated in childhood without lung resection. On the basis of this observation, we suggest that children with this anomaly be treated with simple ligation or that the anomalous vessel be occluded

via transcatheter treatment to avoid the complications of lobectomy.

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