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Case Report
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Fetal Lung Sequestration: An Abnormal Blood Supply

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Abstract

Bronchopulmonary Sequestration (BPS) is a rare congenital anomaly of the lung that receives its blood supply from the systemic circulation. We present a rare case of BPS diagnosed at 24 weeks'-gestation that received its unusual blood supply from the celiac trunk.

Introduction

Congenital masses in the lungs are rare with a prevalence of 1 in 10,000 to 35,000 live births. Bronchopulmonary Sequestration (BPS) presents only 0.15% to 6.4% of all the congenital lung anomalies [1,2]. The difference between fetal BPS to other lung masses is their blood supply evolving from the systemic circulation, usually the aorta, while Congenital Pulmonary Adenoid Malformations (CPAM), and receive their blood supply from the pulmonary circulation. We present a fetus with extra-lobar BPS with an interesting rare blood supply.

Case Presentation

A 25 years-old generally healthy woman was referred to our ultrasound unit for evaluation of fetal lung mass. This was a spontaneous pregnancy, with normal nuchal-translucency, anatomical scan at 15 weeks'-gestation and second trimester biochemical-screening-tests. The level-II anatomical scan at 24 weeks'-gestation revealed an adequate for gestational age fetus, with a homogenous hyperechogenic mass in the left lower lung measuring 44 mm × 22 mm × 31 mm (Figure 1a). Doppler ultrasound demonstrated arterial supply emerging from the abdominal aorta (Figure 1b). The CPAM-volume-ratio (CVR) was 0.6. There were no signs of hydrops. Fetal echocardiography revealed normal anatomy of the heart with dextroposition due to mediastinal shift caused by the lung mass. She received a course of betamethasone and was followed weekly. MRI at 32 weeks'-gestation confirmed the diagnosis (Figure 1c). The size of the lung mass at 35 weeks'-gestation was 50

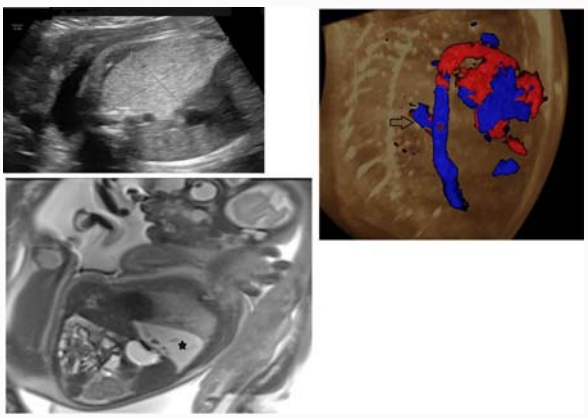

Figure 1a: Axial view of fetal thorax at 26 weeks' gestation demonstrating echogenic structure in the left lower lobe.
Figure 1b: Three dimensional Doppler sagittal ultrasound of the fetal aorta at 26 weeks' gestation, demonstrating an aberrant blood supply from the abdominal portion of the descending aorta (arrow).
Figure 1c: Magnetic resonance imaging of sagittal view of the fetal thorax at 32 weeks' gestation demonstrating a mass in the left lower lung (asterisk).

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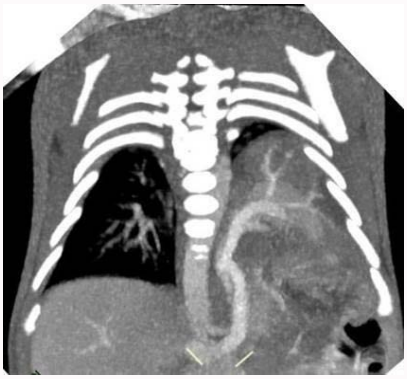

Figure 2: Computerized tomography of the neonate at one month demonstrating a mass in the left lung with blood supply from the celiac trunk.

mm × 35 mm × 20 mm. The CVR was 0.56, with no signs of hydrops. At 38+5 weeks'-gestation mild pericardial and pleural effusion were demonstrated, therefore, labor was induced. Delivery took place at a tertiary center. The patient delivered vaginally a boy of 2729 g who breathed spontaneously with Apgar-score of 9/10. Computerized-tomography-angio revealed extra-lobar lung sequestration located in the lower part of left lung, receiving the blood supply from the celiac-trunk, and venous drainage to the splenic vein and the portal vein (Figure 2). The boy is now seven months old, asymptomatic, and is routinely followed by a pulmonologist. The lung mass did not decrease in size and an operative procedure will take place in the near future.

Discussion

We presented an interesting rare case of extralobar BPS diagnosed at 24 weeks'-gestation, which received its unusual blood supply from the celiac truck and drained into the splenic veins. The pathogenesis of BPS is unknown. In utero airway obstruction in the early pseudoglandular stage (5 to 17 weeks'-gestation), prior to separation of the aortic and pulmonary circulation was suggested as a possible explanation. Another theory is that portion of the developing lung is mechanically separated from the rest by compression from vascular structures, traction by aberrant systemic vessels, or inadequate pulmonary blood flow. Therefore, the prenatal diagnosis of lung masses is usually after 21 weeks'-gestation, and in many cases only following [1-3]. The case we presented was extralobar BPS, located outside the normal lung. These cases comprise 25% of BPS. The hallmark of Bronchopulmonary Sequestration is its anomalous arterial supply, which arises, in most instances, from the aorta. In the case presented here postnatal CT-angiography made the final diagnosis of blood supply from the celiac-truck and venous drainage to the splenic veins.

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