The bridging bronchus and pulmonary artery sling

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Abstract

A 5-month-old male baby was brought to the emergency room with a one-day history of respiratory distress, wheezing, vomiting, and high fever. Wheezing had been present since birth but had recently become more frequent and intense, occurring even at rest. The baby was born full-term by cesarean delivery and had a medical history of gallbladder agenesis, secundum atrial septal defect (ASD), and cryptorchidism.

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The case reports are not required to have an ethics committee review per local policy. The parent of the patient consented to have the case published.

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To the Editor,

A 5-month-old male baby was brought to the emergency room with a one-day history of respiratory distress, wheezing, vomiting, and high fever. Wheezing had been present since birth but had recently become more frequent and intense, occurring even at rest. The baby was born full-term by cesarean delivery and had a medical history of gallbladder agenesis, secundum atrial septal defect (ASD), and cryptorchidism. Physical examination showed prolonged expiration and crackling sounds in the lungs. Vital signs were as follows: fever; 36.4, heart rate; 150, respiratory rate; 55, blood pressure; 90/55, and oxygen saturation on room air; 93%. Laboratory tests showed an elevated c-reactive protein (CRP) level of 5 mg/dL (reference <0.5 mg/dL) and a NT-proBNP (n-terminal pro-brain natriuretic peptide) level of 596 pg/mL (reference <125 pg/mL).

The patient was hospitalized in the pediatric intensive care unit. Chest radiograph showed volume loss and decreased bronchovascular shadows in the left lung, and prominent right pulmonary vascular structures (Figure 1). Contrast-enhanced chest computed tomography (CT) was obtained. Chest CT revealed right pulmonary artery stenosis and a pulmonary artery sling (PAS), in which the left pulmonary artery (LPA) originates from the right pulmonary artery (RPA) and then forms a partial ring around the left main bronchus (LMB). There was a significant compression and stenosis of the LMB due to the PAS (Figure 2A, 2B). Moreover, CT showed left lower lobe pneumonia and an abnormal bronchus that originated from the LMB and extended to the right middle and lower lobes, indicating a bridging bronchus (BB) anomaly (Figure 2C, 2D, and 3). The patient was treated with antibiotics and bronchodilators, and the clinical symptoms regressed. Surgery was recommended for the pulmonary artery sling and bridging bronchus anomaly, but the family said they needed time to consider it, and the patient was discharged.

Bridging bronchus (BB) is a rare congenital anomaly where there is an anomalous bronchus to the right lung arising from the left main bronchus. Patients commonly present with signs and symptoms related to large airway obstruction, such as wheezing, respiratory distress, stridor, and recurrent respiratory tract infections, as in the present case. A total of five types of BB have been defined by Henry et al. according to the origin of the BB and the presence of pseudocarina (1). The present case fulfills the criteria of Type I BB, which is the most common form. BB is a potentially life-threatening condition and usually associated with tracheobronchial anomalies such as congenital long-segment tracheal stenosis and is generally accompanied by congenital cardiovascular anomalies, including left pulmonary artery sling, atrial septal defect (ASD), ventricular septal defect (VSD), tetralogy of Fallot, and coarctation of the aorta (CTS). The most common site of airway stenosis is the LMB, as in the present case (1, 2). Treatment for BB involves surgical intervention, and the timing and extent of surgery depend on the severity of the airway obstruction, the presence of associated anomalies, and the patient's clinical status. The surgical options for bridging bronchus include resection of the anomalous bronchus, reconstruction of the affected airway, and correction of any associated cardiovascular anomalies (1).

Pulmonary artery sling (PAS) is a rare congenital anomaly where the LPA originates from the RPA and wraps around the trachea and/or bronchi (3). This results in a significant compression of the airways, which can lead to respiratory distress and recurrent respiratory infections, as in the present case. PAS is often associated with other congenital anomalies, including tracheobronchomalacia, BB, vascular rings, and cardiovascular defects such as tetralogy of Fallot and patent ductus arteriosus. The diagnosis of PAS can be challenging, and imaging studies such as CT and MRI are typically necessary to confirm the diagnosis (4). Surgical intervention is usually required to relieve the compression of the airways and prevent further respiratory complications. With appropriate management, most patients with pulmonary artery sling can expect a good outcome (3, 4).

In conclusion, this case typically shows the association of BB and PAS in a baby with respiratory distress, which requires immediate management in the pediatric intensive care unit.

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Figure Legends

Figure 1: Anteroposterior chest radiograph showed volume loss and decreased bronchovascular shadows in the left lung, and prominent right pulmonary vascular structures.

Figure 2: a) Contrast-enhanced axial chest computed tomography (CT) image shows right pulmonary artery stenosis (arrowhead) and a pulmonary artery sling (arrows), in which the left pulmonary artery originates from the right pulmonary artery and then forms a partial ring around the left main bronchus. **b)** Axial chest CT image with lung window settings demonstrates a significant compression and stenosis of the left main bronchus (dashed arrow) due to the pulmonary artery sling. Moreover, CT showed left lower lobe pneumonia (*). **c)** Coronal reformatted chest CT image with minimum intensity projection demonstrates an abnormal bronchus that originated from the left main bronchus and extended to the right middle and lower lobes, indicating a bridging bronchus anomaly (arrows). Note the stenosis of the left main bronchus due to the pulmonary artery sling (dashed arrow).

Figure 3: A three-dimensional volume rendering CT image of the posterior view of the tracheobronchial tree shows an abnormal bronchus that originated from the left main bronchus and extended to the right middle and lower lobes, indicating a bridging bronchus anomaly (arrows). Note the stenosis of the left main bronchus due to the pulmonary artery sling (dashed arrow).