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Case Report

A rare duo: Pulmonary arterial sling with a pig bronchus ☆

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ABSTRACT

Pulmonary artery sling is an incomplete vascular ring, where the left pulmonary artery originates from the right pulmonary artery, leading to airway constriction. A tracheal bronchus is an anatomical variation in which an extra bronchus originates from the trachea, frequently resulting in respiratory symptoms or complications. We report a 6-week-old female patient with a pulmonary artery sling coursing around the distal trachea and a concurrent tracheal bronchus.

Introduction

Pulmonary artery sling consists of an abnormal development of the aortic arches in which the left pulmonary artery originates from the right pulmonary artery and courses around the distal trachea, passing between the trachea and the esophagus. Pulmonary artery slings reportedly make up 5% of vascular abnormalities [1]. Even rarer is the coexistence of a pulmonary arterial sling with a tracheal bronchus, also called a "pig bronchus." This congenital variation involves an extra bronchus originating directly from the trachea. The term "pig bronchus" is used because this variation is more commonly observed in pigs and certain animals. Identifying a tracheal bronchus is important because when present in humans, it can lead to respiratory problems like recurrent infections, chronic cough, aspiration, and breathing difficulties [2].

Presenting symptoms of a pulmonary arterial sling with a tracheal bronchus may include respiratory distress and stridor due to compression of the trachea, as well as signs of respiratory tract infection or recurrent pneumonia [2]. Initial evaluation of a suspected vascular ring can include a barium esophagogram, echocardiogram, or bronchoscopy (Fig. 1). Diagnosis is confirmed by CT angiography or magnetic resonance angiography (MRI angiography), which provides surgeons with the necessary information for preoperative planning. The clinical

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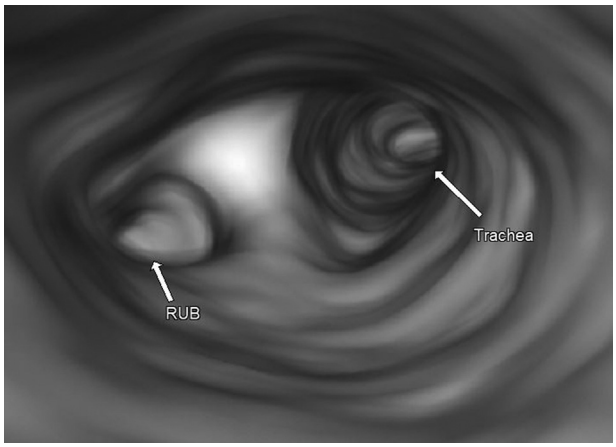

Fig. 1 – Volume rendered virtual bronchoscopy showing right upper lobe tracheal bronchus.

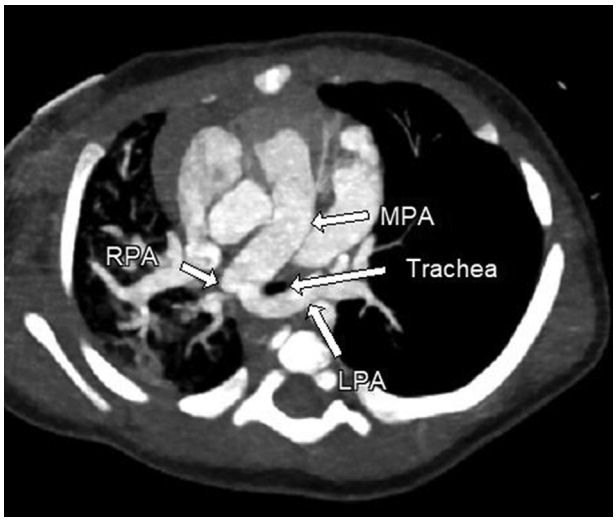

Fig. 2 – Axial image from a chest CTA demonstrates pulmonary artery sling, with the left pulmonary artery arising from the proximal right pulmonary artery and wrapping around the distal trachea.

case of an infant with a pulmonary arterial sling and tracheal bronchus is reported.

Case presentation

A 6-week-old female presented to Banner Desert Medical Center for evaluation of respiratory distress and weight loss. On physical exam, a significant murmur was heard that had not previously been noted on prior exams. Initial echocardiogram revealed a left pulmonary arterial sling, prominent patent foramen ovale, and a moderate size patent ductus arteriosus (PDA) with continuous left to right shunting. Subsequent CT angiogram confirmed a pulmonary sling, with the left pulmonary artery arising from the distal right pulmonary artery and wrapping around the distal trachea (Fig. 2). Left proximal

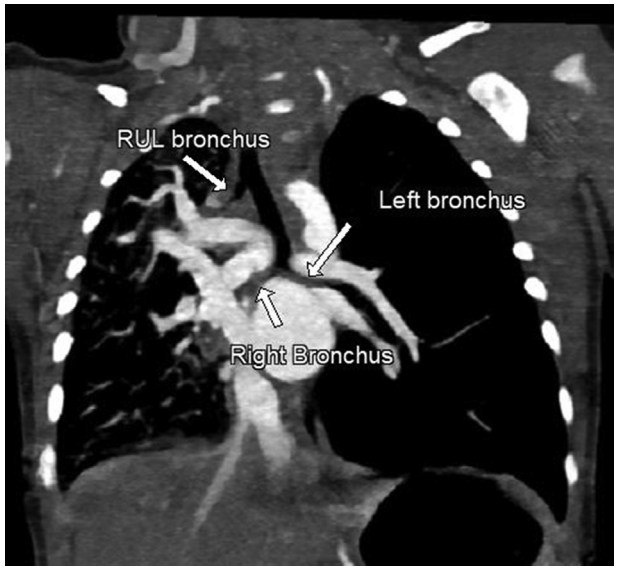

Fig. 3 – Coronal image from a chest CTA showing right upper lobe tracheal bronchus with right and left main stem bronchi seen inferiorly.

mainstem bronchus obstruction, consistent with left-sided air trapping, was observed. Moderate-sized patent ductus arteriosus (PDA) and a notable patent foramen ovale (PFO) were present. Furthermore, a right upper lobe tracheal bronchus (pig bronchus) was identified (Fig. 3). At 7-months-old the patient underwent left pulmonary artery sling and PDA repair with re-implantation of the left pulmonary artery into the main pulmonary artery anterior to the trachea and PDA division/ligation. She developed postoperative left pulmonary artery stenosis and underwent successful balloon angioplasty. Unfortunately, due to the significant left pulmonary artery hypoplasia she developed recurrent left pulmonary artery stenosis.

Discussion

The unique co-occurrence of a pulmonary artery sling and a tracheal bronchus in our case highlights the complexity of respiratory anatomical variations and prompts a comprehensive discussion of diagnostic challenges, clinical implications, and management for this rare anatomical presentation. CT angiography is the gold standard technique for diagnostic evaluation and surgical planning because it allows for detailed evaluation of the airway and vasculature. Echocardiogram may be a helpful imaging tool; however, it has lower sensitivity and would not detect a tracheobronchial tree abnormality [3]. In addition to CT imaging, 3D reconstruction techniques play a pivotal role in comprehensively visualizing the intricate anatomy associated with pulmonary arterial sling and tracheal bronchus anomalies, offering valuable insights that can aid in diagnosis and inform optimal patient management strategies (Fig. 4).

Clinical presentation depends on the degree of compression that these vascular structures produce on the airway

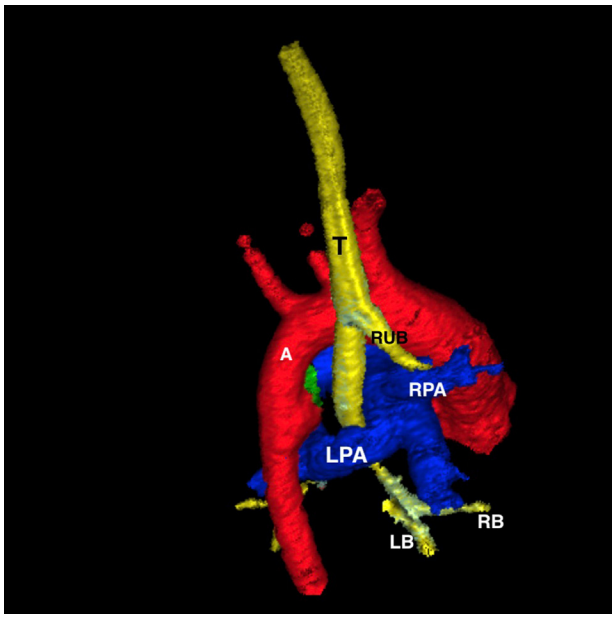

Fig. 4 – Volume rendered color coded 3D model from chest CTA showing the tracheal bronchus and pulmonary sling.

and/or the esophagus. Patients typically present with respiratory symptoms in the first year of life. Older children tend to present with esophageal symptoms. The presence of a tracheal bronchus can have clinical implications, especially if it causes respiratory symptoms or if it is inadvertently intubated during procedures such as bronchoscopy or endotracheal intubation. Detecting this condition in children is particularly important because of their smaller airways, higher vulnerability to respiratory issues, and the potential need for surgical correction if complications arise [4].

Pulmonary artery slings are frequently associated with cardiovascular malformations and tracheobronchial anomalies [5]. Treatment for pulmonary artery slings remains surgical and involves addressing the anomalous course of the left pulmonary artery to relieve its compression on the trachea. Symptomatic patients with these complex lesions have a high mortality rate without surgical intervention. Median sternotomy is the best approach for repair because it allows concomitant repair of intracardiac defects [6]. Surgical complications include bronchial leak, infection, right heart failure, and respiratory failure.

Conclusion

Pulmonary arterial sling with an associated tracheal bronchus is a rare and complex congenital anomaly that requires the role of imaging for diagnosis. The integration of CT imaging and advanced 3D reconstruction techniques offers a powerful synergy, enabling identification of these rare anomalies and visualization of their intricate anatomical relationships, facilitating more informed clinical decisions.

Patient consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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