

Case Report

Partial lung resection of supernumerary tracheal bronchus combined with pulmonary artery sling in an adult: Report of a Case

Running Head: lung resection of supernumerary tracheal bronchus

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Abstract

An adult case of pulmonary resection for repeated infections in a supernumerary tracheal bronchus combined with a pulmonary artery sling is reported. A 33-year-old woman with a pulmonary artery sling was referred for recurrent lung infections. Chest computed tomography showed the left pulmonary artery arising from the right pulmonary artery and coursing posterior to the trachea. The lung parenchyma connected to the tracheal bronchus showed dense opacity and traction bronchiectasis. Partial pulmonary resection was performed with an ultrasonically activated scalpel after the tracheal bronchus was auto-sutured. The patient's postoperative course was uneventful, and she is now in good condition.

Introduction

Pulmonary artery sling (PAS) is a rare vascular anomaly in which the left pulmonary artery arises from the right pulmonary artery usually detected in infants. It is sometimes combined with tracheal anomalies and causes compression of the lower trachea [1]. However, there are few reports about PAS detected in adulthood. Herein, we report a case of pulmonary resection for repeated infections in a supernumerary tracheal bronchus combined with a pulmonary artery sling in an adult.

Case

A 33-year-old woman with dry cough and a low-grade fever visited a regional hospital. She was treated with antibiotics and recovered fully. She had already been diagnosed as having a left pulmonary artery sling in her childhood, but she remained untreated because she had refused treatment. She was referred to our department for further examination and evaluation for surgical treatment. Chest X-ray showed a slight opacity in the upper mediastinum. 3D and enhanced chest computed tomography confirmed the left pulmonary artery arising from the right pulmonary artery and coursing posterior to the narrowed trachea towards the left thorax (Fig. 1A, 1B). Moreover, a right supernumerary tracheal bronchus was identified above the carina, and ground-glass opacity and traction bronchiectasis were seen (Fig. 2). No other cardiovascular anomalies were identified on echocardiography. Her pulmonary function tests showed that the vital capacity (VC), percent of vital capacity (%VC), forced expiratory volume per 1 second (FEV1.0), and forced expiratory volume 1% (FEV1.0%) were 2870 mL, 95.0%, 1990 mL, and 69.1%, respectively. Peak expiratory flow was 3.6 L/s, which was 44.3% predicted. Arterial blood gas analysis showed that the arterial oxygen (PaO₂) and carbon dioxide (PaCO₂) pressures on room air were 84.7 mmHg and 37.8 mmHg, respectively. On

bronchoscopy, the tracheal bronchus was located in the six o'clock position (membranous portion), a small amount of foamy sputum was suctioned (Fig. 3), and a flattened carina was seen. She had a history of recurrent pneumonia in her childhood, assumed to be related to this tracheal abnormality. Therefore, it was decided to perform a pulmonary resection for the infected lesion connected to the tracheal bronchus. Surgical repair of the pulmonary artery sling and tracheal stenosis was omitted in this operation.

Anterolateral thoracotomy was done at the fourth intercostal space because one-lung ventilation could not be performed by either a double lumen tube or a blocker tube due to the tracheal stenosis and lower located bifurcation. A spiral tube was inserted beyond the orifice of the tracheal bronchus. A mild fibrous adhesion was seen around the upper mediastinal pleura. The left pulmonary artery was identified arising from the right pulmonary artery, and the right upper truncus of the pulmonary artery was taped. These main vessels were not running into the lung parenchyma connecting to the tracheal bronchus. The tracheal bronchus was easily identified below the azygos vein and auto-sutured. Partial pulmonary resection of the infectious focus was performed with both electrocautery and an ultrasonically activated scalpel after the borderline between inflated lung and deflated lung was identified. Fig. 4 was the schema after this operation. The operation time was 227 minutes, and the total amount of bleeding was 130 mL. The patient's postoperative course was uneventful, and she was discharged on the 8th postoperative day. She remains in good condition with no symptoms.

Discussion

A case of pulmonary resection for repeated infections in a tracheal bronchial anomaly combined with a left pulmonary artery sling (PAS) in an adult was described.

PAS is a rare vascular anomaly in which the left pulmonary artery arises from the right pulmonary artery. It is sometimes combined with tracheal anomalies and causes compression of

the lower trachea [1]. Most PAS cases are identified with severe respiratory distress in infants. Speggorin et al. [2] reported 84 pediatric cases that underwent slide tracheoplasty for long-segment congenital tracheal stenosis. A tracheal right upper lobe bronchus was present in 10 patients (11.9%), and PAS was observed in 8 patients. No deaths occurred in patients with right upper lobe bronchus anatomy. Moreover, Huang et al. [3] reported that approximately 60% of the patients with PAS underwent tracheoplasty, and a tracheal diameter of less than 3 mm was associated with poor outcomes. In the present case, the patient's peak expiratory flow was significantly decreased (only 44.3% predicted), which suggested stenosis of the respiratory tract. However, the 5.3-mm-diameter bronchoscope was easily passed into the lobar bronchus, and there were no findings related to the tracheal rings. Yong et al. [1] reported surgical management of pulmonary artery sling in 21 children. Operative mortality was 14.3%, and it was determined by the need for tracheal surgery. According to these reports, surgery for supernumerary tracheal bronchus was performed safely unless tracheal surgery was necessary. Unlike in infants, most adults are asymptomatic or have mild symptoms that could be attributed to a PAS [4]. Though LaBelle et al. [4] reported successful surgical repair of PAS in an adult, Procacci et al. [5] reviewed 17 cases of adults diagnosed with PAS and found that none had undergone surgical repair.

A tracheal bronchus is any airway that emerges from the lateral trachea wall and is present in 0.1-2% for right and 0.3-1% for left tracheal bronchus. The main embryogenic hypotheses are the reduction, migration, and selection theories [6]. According to the report of Maeda et al [7] intra-pulmonary arteries arises from primitive vessels associated with budding airways in the development. However, we could not detect the obvious vessels running into the lung parenchyma connecting to the tracheal bronchus. The pathogenesis of anomalous bronchial development remains somewhat controversial [6].

In the present case, curative operation such as re-implantation of the PAS and tracheoplasty, which requires cardiopulmonary bypass, was avoided. Furthermore, an attempt was made to

preserve pulmonary function as much as possible, because the patient was young and had no other symptoms during her daily life except for the repeated infectious episodes. Although her infectious episodes may have been related to the tracheal stenosis rather than the tracheal bronchus, she had no history of bronchial asthma or wheezing, and no limitation of daily life. Thus, only partial pulmonary resection related to the tracheal bronchus was performed. However, further follow-up is needed with respect to the outcome of this operation and the clinical course of tracheal stenosis and PAS.

Needless to say, it is important for surgeons to identify the left pulmonary artery seen in the hilum and not to injure it during this surgery. Moreover, since one-lung ventilation was not possible due to the tracheal stenosis, thoracotomy was needed. However, Wiser et al. [6] reported successful isolation of the right lung by a combined technique involving a Fogarty catheter and an endobronchial blocker. Video-assisted thoracic surgery is possible and desirable with this technique for selected cases. To the best of our knowledge, there have been no reports about pulmonary resection of bronchial anomalies combined with PAS.

Conclusion

This is the first case report of pulmonary resection for repeated infections in a supernumerary tracheal bronchus combined with a pulmonary artery sling in an adult. Pulmonary resection is necessary when repeated infections occur in the tracheal bronchus.

Disclosures and Freedom of Investigation

The authors have no personal conflicts of interest and have received no outside support for this research.

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Figure legends

Figure 1 (A-B). Three dimensional enhanced CT shows a supernumerary tracheal bronchus (arrow) and the left pulmonary artery (open arrow) arising from the right pulmonary artery running behind the narrowing trachea.

Figure 2. Chest CT shows ground-glass opacity with traction bronchiectasis at the medial side of the right upper lobe. This opacity is connected to the tracheal bronchus.

Figure 3. Bronchoscopic findings. Tracheal bronchus is located in the membranous portion (arrowed).

Figure 4. The operative schema. Right upper truncus artery (arrow) originated from PAS, and superior pulmonary vein (open arrow) was behind the r-PA.

AV: azygous vein, s-TB: stump of tracheal bronchus, PAS: pulmonary artery sling, r-PA: right pulmonary artery, m-PV: middle pulmonary vein.

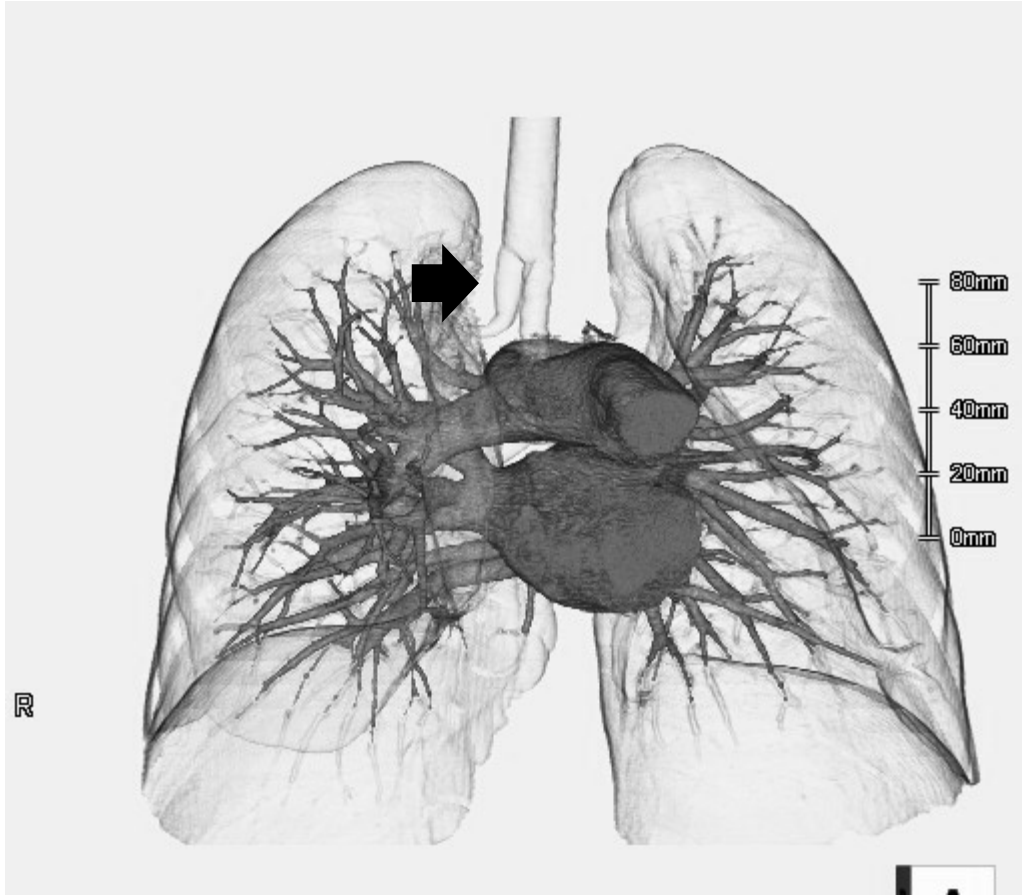


Fig. 1A.

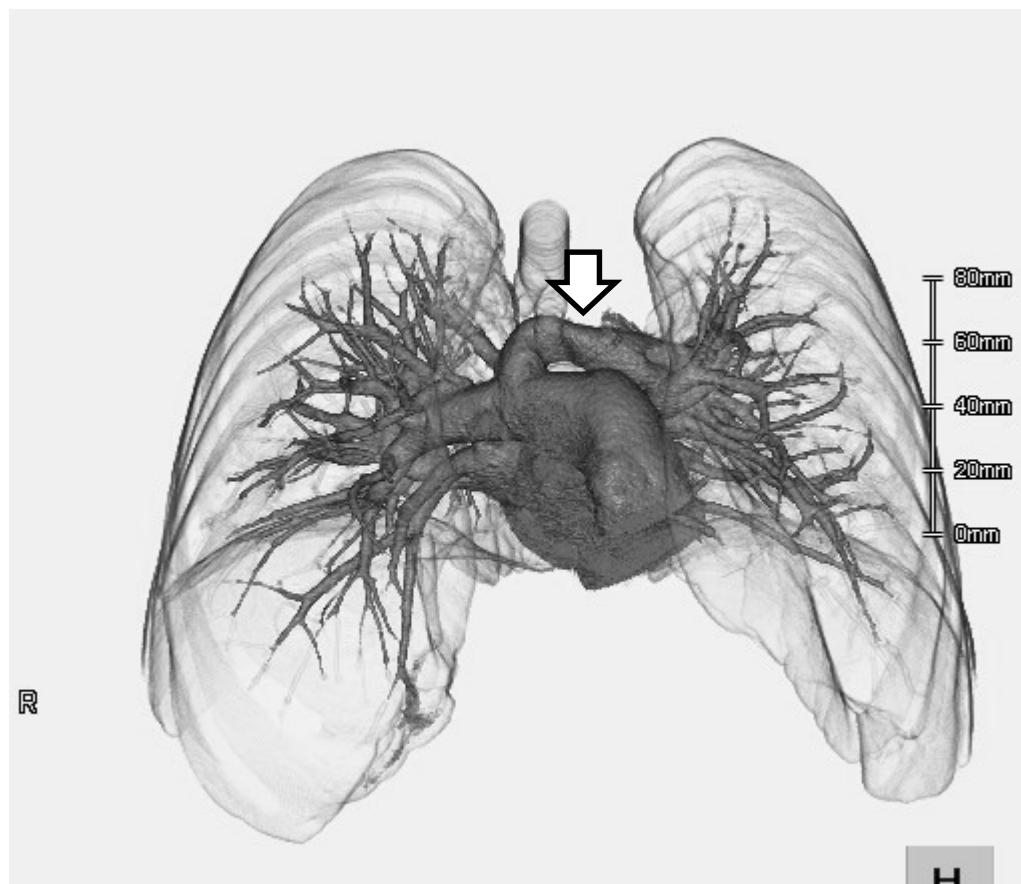


Fig. 1B.

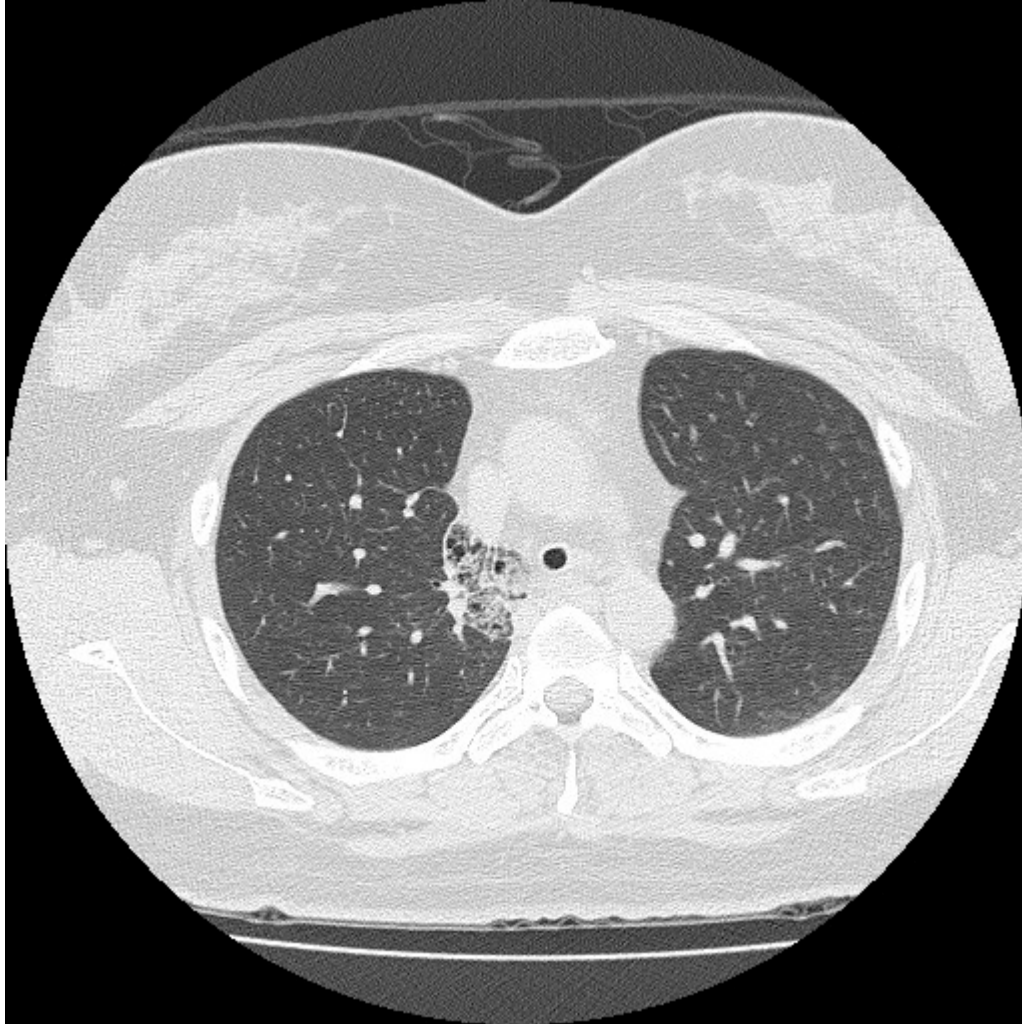


Fig. 2.

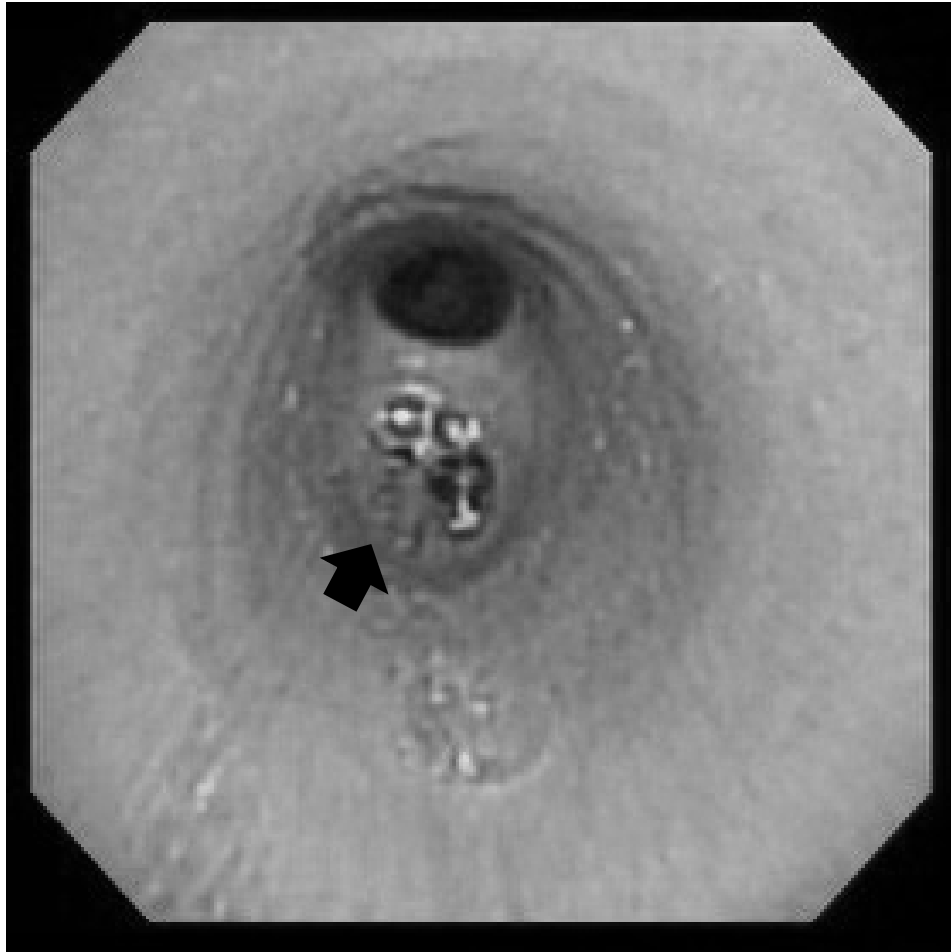


Fig. 3.

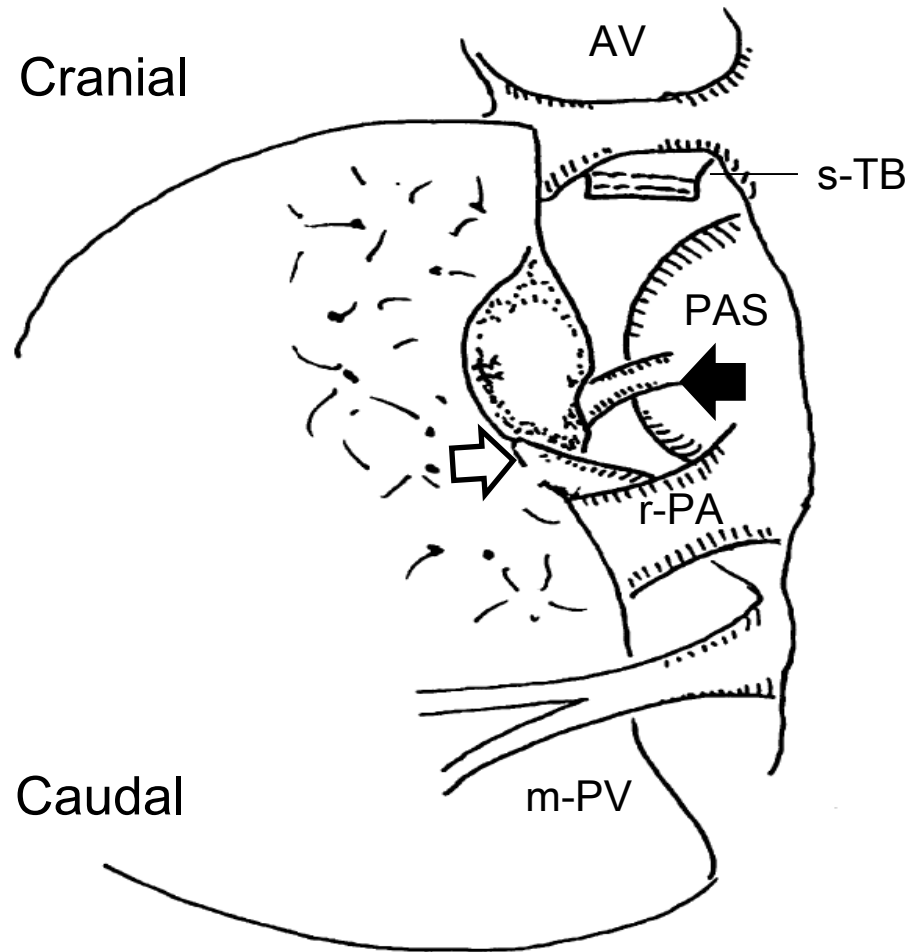


Fig. 4.