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Case Report

Asymptomatic discovery of a rare congenital vascular anomaly in a 33-year-old male: a case of Pulmonary Sling Syndrome

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ABSTRACT

Pulmonary sling (PS) syndrome is a rare congenital vascular anomaly characterized by the anomalous origin of the left pulmonary artery (LPA) from the right pulmonary artery (RPA). While it typically presents in infancy with severe respiratory distress, its diagnosis in adulthood is exceedingly rare and often incidental. A 33-year-old male presented with acute, severe right-sided pleuritic chest pain. Initial suspicion was pulmonary embolism (PE); however, laboratory markers (D-dimer, troponin) were normal. Pulmonary CT angiography ruled out PE but revealed an incidental pulmonary sling, where the LPA originated from the RPA and coursed between the trachea and esophagus. A 10 mm pleural effusion was noted. The patient was diagnosed with idiopathic pleurisy, treated with anti-inflammatory agents, and symptoms resolved. This case underscores the possibility of PS remaining asymptomatic until the fourth decade of life. Clinicians should be aware of such vascular variants, as they may have significant implications for airway management and thoracic surgery.

Keywords: pulmonary artery, vascular ring, incidental findings

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Introduction

Pulmonary sling syndrome, first described by Glaevecke and Doehle in 1897, is a unique form of vascular ring [1]. Unlike other vascular rings that encircle both the trachea and esophagus, the pulmonary sling specifically involves the anomalous origin of the left pulmonary artery (LPA) from the posterior aspect of the right pulmonary artery (RPA). As the LPA courses toward the left hilum, it passes between the distal trachea and the anterior esophagus, creating a "sling" or "lasso" effect that can lead to mechanical compression of these structures [2].

The incidence of this anomaly is estimated to be approximately 1 in 17,000 live births. Due to the high frequency of associated tracheobronchial tree malformations, such as complete tracheal rings (O-shaped rings) and tracheal stenosis, most patients present with life-threatening respiratory distress, stridor, or recurrent pneumonia shortly after birth [3]. However, in a very small subset of patients where the airway is structurally robust or the compression is mild, the condition may remain clinically silent. The literature regarding adult-onset or incidentally discovered pulmonary sling is sparse, making each documented adult case vital for understanding the natural history of this congenital malformation [4].

Case Report

A 33-year-old male with no prior significant medical history presented to the emergency department with acute, stabbing right-sided chest pain. The pain was exacerbated by deep inspiration and physical movement, typical of pleuritic irritation. On physical examination, the patient was hemodynamically stable (BP: 120/80 mmHg, HR: 86 bpm, SpO₂: 97%). Lung auscultation revealed equal breath sounds without rales or rhonchi.

Laboratory workup showed a normal D-dimer level and negative cardiac troponins. C-reactive protein (CRP) was initially 4.1 mg/L. Despite the low probability based on the Wells criteria and normal D-dimer, the severity and persistence of the pain necessitated a pulmonary CT angiography (CTA) to definitively rule out pulmonary embolism (PE).

The CTA excluded PE but revealed an incidental pulmonary sling. The LPA was observed arising from the RPA and coursing posteriorly between the trachea and esophagus, just above the carina, toward the left lung (Figures 1-3). A concurrent 10 mm right-sided pleural effusion was identified. The patient was monitored; on the second day, his CRP rose to 10.5 mg/L, supporting the diagnosis of idiopathic pleurisy. Treatment with non-steroidal anti-inflammatory drugs (NSAIDs) was initiated. At the one-week follow-up, the CRP had decreased to 4.9 mg/L, and the patient's symptoms had

completely resolved. The pulmonary sling was documented as an incidental, asymptomatic finding.

Following the initial identification of the anomaly on CT, a cardiovascular surgery consultation was performed to assess the potential for surgical intervention. A comprehensive evaluation, including axial MIP and 3D reconstructions, revealed no evidence of critical hemodynamic compromise or hemodynamically significant tracheobronchial stenosis. Consequently, in the absence of symptoms, a conservative strategy involving regular clinical and radiological monitoring was adopted by the multidisciplinary team. Written informed consent for publication was obtained from the patient.

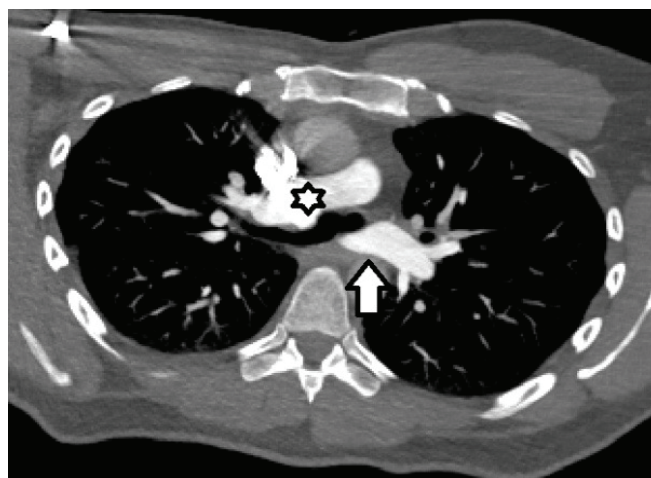


Figure 1. Axial CT angiography image demonstrating the left pulmonary artery (LPA) originating from the right pulmonary artery (RPA) and its course posterior to the trachea (asterisk: RPA, arrow: LPA).

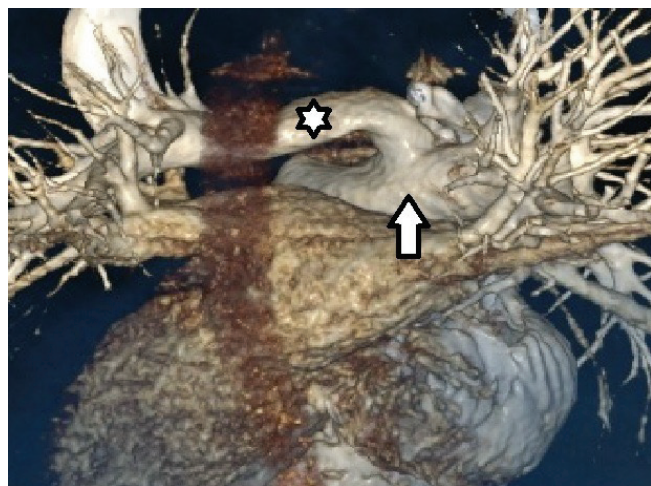


Figure 2. Posterior view of the three-dimensional volume-rendered CT reconstruction. The image demonstrates the heart, left atrium (black arrowhead), and pulmonary veins (white arrowheads). The right main pulmonary artery is visualized (black arrow), along with the anomalous left pulmonary artery (asterisk), which originates from the right pulmonary artery and follows a retro-cardiac course. The esophagus is observed as a columnar structure positioned anterior to the anomalous left pulmonary artery (white arrows).



Figure 3. Axial Maximum Intensity Projection (MIP) image showing the anomalous left pulmonary artery (white asterisk) arising from the right pulmonary artery (black asterisk) and passing between the trachea (white arrow) and the esophagus (black arrow), confirming the diagnosis of pulmonary artery sling syndrome.

Discussion

The embryological development of the pulmonary arteries is a complex process occurring between the 4th and 7th weeks of gestation. Normally, the pulmonary arteries develop from the proximal part of the sixth branchial arches. A pulmonary sling occurs when the left sixth arch fails to develop correctly, and a persistent postbranchial vessel (capillary plexus) from the right side connects to the left lung bud [5]. This abnormal connection forces the developing left pulmonary artery to originate from the right pulmonary artery, crossing the midline to reach the left lung.

Pulmonary slings are classified into two main types based on the Landing and Wells system [6]:

Type 1 (Classic): The LPA arises from the RPA, and the bifurcation of the trachea (carina) is at the normal level (T4-T5). The sling compresses the right main bronchus and the lower trachea.

Type 2 (Complex): This is more common and involves an abnormally low carina (T6 level) and "T-shaped" bronchi. This type is frequently associated with "bridging bronchus" and long-segment tracheal stenosis due to complete O-shaped rings.

Our case likely represents a Type 1 variant, given the absence of chronic respiratory symptoms and the normal carinal position observed on imaging.

While the classic presentation involves 'the ring-sling complex' in neonates, adult cases are often discov-

ered incidentally during routine lung screenings or during the evaluation of unrelated clinical symptoms [7,8]. The lack of symptoms in adults is generally attributed to the absence of intrinsic tracheal abnormalities. In our patient, the discovery was purely incidental during an evaluation for chest pain. This highlights that while the "sling" is anatomically present, it may not always be physiologically obstructive.

In symptomatic infants, the gold standard of treatment is surgical reimplantation. This involves detaching the LPA from the RPA and anastomosing it to the main pulmonary trunk, anterior to the trachea [9]. If tracheal stenosis is present, simultaneous slide tracheoplasty is performed. In contrast, for asymptomatic adults, the management is controversial. Most experts recommend a conservative "wait and watch" approach, as the risks of major thoracic surgery usually outweigh the benefits in the absence of airway compromise [10]. However, its presence should be noted, as it can complicate endotracheal intubation, transbronchial biopsy, or future mediastinal surgeries [11].

Although PAS is classically associated with severe respiratory distress in infancy, this case demonstrates that the anomaly can remain clinically silent until the fourth decade of life. We hypothesize that the lack of intrinsic tracheal abnormalities and a relatively preserved airway diameter provided a compensatory mechanism that prevented early clinical manifestation.

In conclusion, pulmonary sling syndrome in adults is a rare but important diagnostic entity. This case demonstrates that the anomaly can remain silent for over three decades. CT angiography remains the definitive tool for diagnosis; however, its incidental finding should prompt a comprehensive evaluation of the airway using both advanced radiological imaging and, when clinically indicated, bronchoscopy to exclude underlying tracheobronchial malformations.

Declaration of conflicting interests

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Authors' contribution

All authors contributed to the conception, data collection, writing, and final approval of the manuscript.

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