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Pulmonary sequestration versus pseudo sequestration: Diagnostic and pathologic correlation in a complex case

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Abstract:
Pulmonary sequestration is a rare anomaly involving non-functional lung tissue. A 68-year-old patient presented with recurrent respiratory infections underwent contrast-enhanced CT thorax, revealing an encapsulated rounded opacity in posterior basal segment of left lower lobe with communication to the segmental bronchi. Left lower lobectomy was performed and histopathology showed congestion, fibrosis, abscess formation and cavities containing actinomycotic colonies and fungal balls. This case highlights importance of combining imaging and histopathological findings for accurate diagnosis and management of complex pulmonary conditions. In our case, inflammatory changes resulted in the narrowing of the vessel and it also complicated radiological interpretation. Thus, data shows the need for the management and proper treatment planning is required to prevent life threatening outcomes.

Keywords: Pulmonary sequestration, tracheobronchial tree, systemic arterial, supply, Lobectomy

Background:
Pulmonary sequestration is a rare congenital anomaly characterized by non-functional lung tissue that lacks normal communication with the tracheobronchial tree and is supplied by an aberrant systemic artery **[1]**. Communication between a sequestrated lung segment and the tracheobronchial tree is extremely rare and not typically associated with classic pulmonary sequestration. Therefore, it is of interest to describe the case of pulmonary sequestration with tracheobronchial tree communication, highlighting its clinical presentation, diagnostic challenges and surgical management.

Case report:
This case involves a 68-year-old male who presented with recurrent lower respiratory tract infections over several months. The patient reported symptoms of intermittent cough, fever and dyspnea, which were unresponsive to standard antibiotic therapy. There was no significant past medical or family history of respiratory disease. Previous imaging evaluation documented as pseudosequesteration, as scan failed to identify vascular supply from aorta. On current admission physical examination elicited, decreased breath sounds in the left lower lung zone, along with dullness on percussion. Routine laboratory investigations were unremarkable, except for mildly elevated inflammatory markers. Chest X-ray suggested opacity in the left lower lobe. To further evaluate, a contrast-enhanced computed tomography (CECT) scan of the thorax was performed, revealing an encapsulated, rounded opacity in the posterior basal segment of left lower lobe **(Figure 1)** with tiny systemic arterial supply (**Figure 2**) from descending thoracic aorta, adjacent pleural thickening and communication to the segmental bronchi (**Figure 3**). In view these features, main diagnosis of pulmonary intralobar sequestration with super added infection, differential diagnoses of pseudo pulmonary sequestration were considered. Based on these findings the patient was prepared for surgical intervention. A left lower lobectomy was performed to remove the affected lobe. Intraoperative findings confirmed intralobar pulmonary sequestration involving the posterior, lateral and anterior basal segment, with multiple systemic arterial supplies (**Figure 4, 5**). The post-operative course was stable initially, within 2 days patient deteriorated, undergone respiratory failure. Histopathological examination of the resected specimen revealed significant congestion and fibrosis of the lung parenchyma. Additionally, there were areas of abscess formation and cavities containing actinomycotic colonies and fungal balls, surrounded by dilated alveoli with neutrophilici nfiltrates and fibrosis which were unexpected findings. These findings confirmed the diagnosis of a complicated pulmonary sequestration with secondary infections (**Figure 6-9**). The patient was treated with a prolonged course of antibiotic therapy, even with immense supportive care following surgery, patient succumbed.

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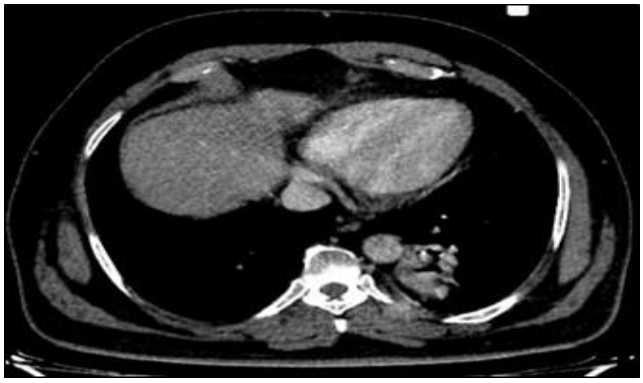

Figure 1: CECT Thorax – demonstrating heterogeneous lobulated lesion in the posterior basal segment of left lower lobe

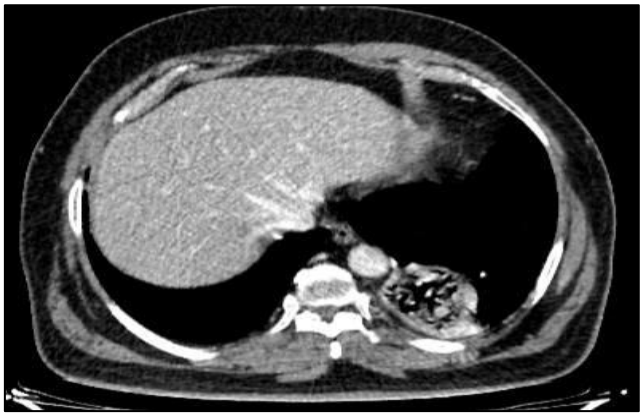

Figure 2: CECT Thorax – demonstrating tiny arterial feeders from descending thoracic aorta, which is difficult to identify

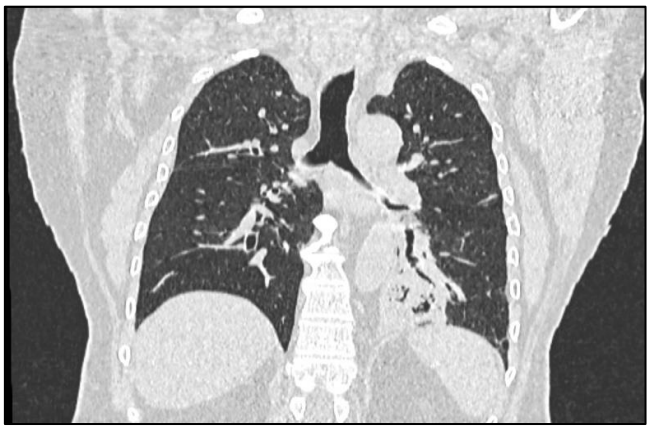

Figure 3: Demonstrating communication to the lower lobe bronchus

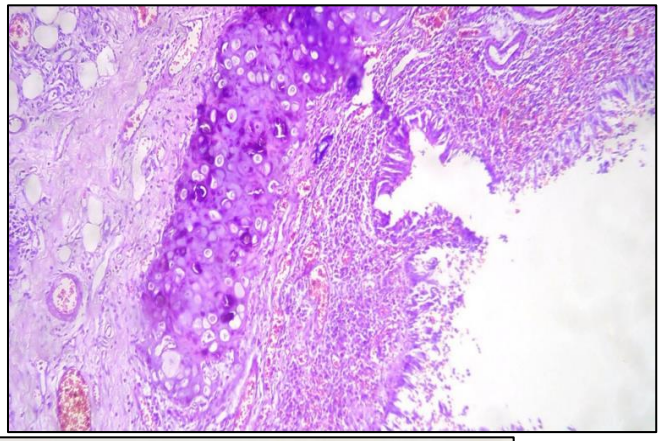

Figure 4: Gross specimen showing granular material as content

Figure 5: Gross specimen showing granular material, forceps used to expose contents

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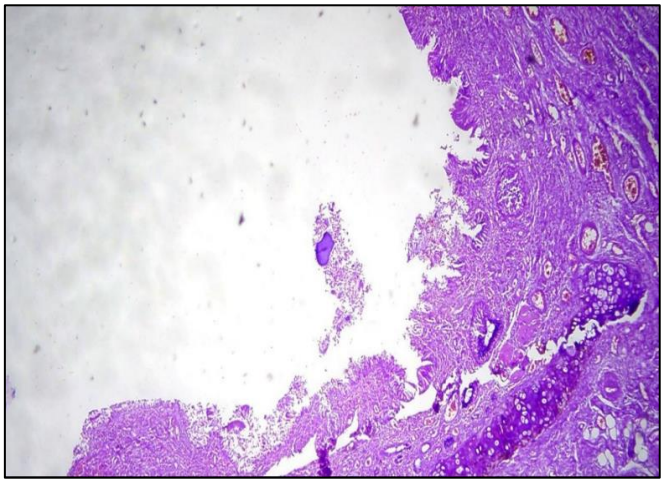

Figure 6: Cavity lined by columnar epithelium, actinomycosis in the cavity (low magnification)

Figure 7: Cavity lined by columnar epithelium, actinomycosis in the cavity (high magnification)

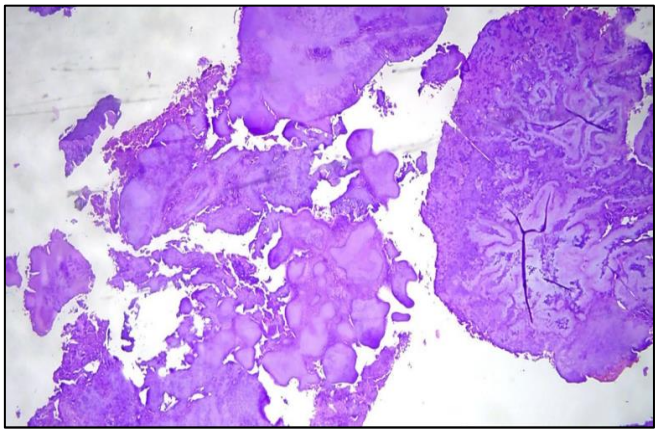

Figure 8: Actinomycosis colonies in the cavity, fungal colonies in Grocott methenamine silver stain (low magnification)

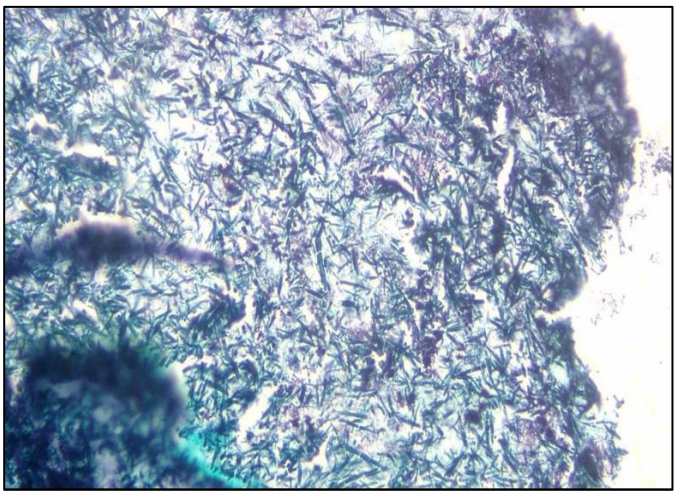

Figure 9: Actinomycosis colonies in the cavity, fungal colonies in Grocott methenamine silver stain (high magnification)

Discussion:

Pulmonary sequestration is a rare congenital anomaly involving non-functional lung tissue that lacks a normal connection to the tracheobronchial tree and often receives blood supply from the systemic circulation **[2]**. It accounts for upto 6% of congenital pulmonary malformations **[3]**. It involves lung tissue that is isolated from then normal bronchopulmonary tree and receives arterial blood supply from systemic circulation, typically the thoracic or abdominal aorta. Pulmonary sequestration is classified into two types **[4].** Intralobar sequestration (ILS): Located within the normal lung parenchyma, it is the more common type. Extralobar sequestration (ELS): Found outside the normal lung and usually encapsulated by its pleura. Pulmonary sequestration may present with a wide spectrum of symptoms, ranging from asymptomatic cases to recurrent pulmonary infections, chronic cough, hemoptysis, or respiratory distress **[5].** In some cases, it is detected incidentally on imaging studies. Secondary infections can complicate the condition, leading to abscess formation and more severe symptoms, as observed in this case **[6].** The diagnosis of pulmonary sequestration relies heavily on imaging studies. CECT thorax is the primary modality, as it provides detailed information on the location, extent and vascular supply of the sequestered lung tissue **[7].** The hallmark of pulmonary sequestration on imaging is the demonstration of lung tissue that receives an anomalous vascular supply and does not communicate with the rest of the tracheobronchial tree. In this case, communication with the tracheobronchial tree was visualized, which is an uncommon finding and presented a diagnostic challenge. However, histopathological analysis remains the definitive diagnostic tool, particularly in cases with unusual presentations or complications **[8-10].** In this case, histopathology revealed unexpected secondary infections, including actinomycotic colonies and fungal balls. Actinomycosis is a rare, chronic bacterial infection caused by *Actinomyces species*, often associated with poor oral hygiene or aspiration **[11].** Fungal balls, most commonly due to *Aspergillus species*, occur in pre-existing lung cavities, as seen in this case. The coexistence of actinomycotic colonies and fungal balls within a sequestrated lobe is rare and underscores the susceptibility of sequestrated lung tissue to infections due to impaired drainage and poor vascularisation **[12, 13].** The management of pulmonary sequestration typically involves surgical resection, especially in symptomatic cases or those with recurrent infections. Lobectomy or segmentectomy is the procedure of choice, as it provides definitive treatment by removing these quartered lung tissue and preventing further complications **[14].** In this case, a left lower lobectomy was performed and the patient's recovery was uneventful. Post-operative antimicrobial therapy targeting the identified pathogens was essential to prevent residual or disseminated infection. Our case is an interesting scenario as in our case; the initial imaging did not identify the aberrant systemic vessel which was arising from the aorta. The reason believed is likely to

be superimposed infection also it's associated inflammatory changes, especially with fungal etiology; this may have led to narrowing of the vessel, which also obscured its visualization in the diagnosis. These factors will complicate radiological assessment and also interferes with accurate diagnosis. This case illustrates the complexity of pulmonary sequestration, particularly when an atypical feature, such as the communication with tracheobronchial tree possibly due to secondary infections, is present **[15].** It emphasizes the role of imaging and histopathology in diagnosis and the importance of surgical and medical management in achieving favourable outcomes. Early recognition and intervention are crucial to prevent complications and improve quality of life in affected patients. This report contributes to the understanding of pulmonary sequestration and its potential complications, adding valuable insights for clinicians managing similar cases. This case illustrates how severe infection can mask critical anatomical details, such as the aberrant systemic vessel and create significant diagnostic challenges. There was difficult in the identification of the vessel as there was narrowing and this emphasized on how easily vital findings cannot be seen in such circumstances. Equally important, this experience highlights the necessity of aggressive and timely management when pulmonary sequestration is complicated by high-grade infection. If proper diagnosis and treatment is not done, the survival will be at risk. These patients require the highest level of clinical vigilance, multidisciplinary collaboration and a carefully planned treatment strategy to anticipate complications and improve outcomes.

Conclusion:

Pulmonary sequestration can present with atypical features. While the absence of communication with tracheobronchial is a hallmark of pulmonary sequestration, its presence on imaging, as seen in this case, underscores the variability in presentation. Clinicians must consider pulmonary sequestration in patients with recurrent respiratory infections and use histopathological examination to confirm the diagnosis when imaging findings are inconclusive.

Declaration of patient consent:

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of Interest:

There are no conflicts of interest.

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