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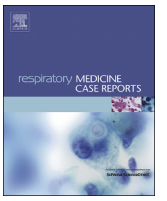

Case Report

A unique case of intralobar pulmonary sequestration in an elderly patient: A case report

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ABSTRACT

Pulmonary sequestration (PS) is a rare congenital anomaly characterized by non-functional lung tissue receiving blood supply from an abnormal source. PS is typically diagnosed in young individuals but is uncommon in the elderly. This abstract describes a case of PS in a 62-year-old male patient presenting with recurrent fever, chronic cough, and difficulty breathing. Imaging revealed abnormal lung tissue disconnected from the bronchial tree, with blood supply from the descending thoracic aorta. Surgical intervention successfully treated the condition. The case emphasizes the need to consider PS as a possible diagnosis, even in older patients, and suggests further research into its possible etiologies.

1. Background

Pulmonary sequestration (PS) is a medical condition characterized by the presence of an anomalous segment or lobe of lung tissue that exhibits abnormal development and structure. This tissue is disconnected from the normal respiratory system and receives its blood supply from a separate, anomalous source, rather than the typical pulmonary circulation. Consequently, this nonfunctional tissue does not contribute to the normal respiratory function of the lung [1]. PS is a congenital anomaly that originates from the primitive foregut during fetal development. The prevailing explanation for this condition suggests the formation of an additional supernumerary lung bud beneath the normal lung bud [2]. Incidence of pulmonary sequestration is relatively rare, accounting for approximately 1–6% of all congenital lung abnormalities, and it may go undetected during prenatal stages or early childhood [3]. The term "pulmonary sequestration," also known as bronchopulmonary sequestration, was first introduced by Pryce in 1946 [4]. Based on the positioning of the abnormal segmental lung tissue relative to the pleura, pulmonary sequestration can be categorized into two distinct types: intralobar and extralobar [5]. In adults, intralobar sequestration is often incidentally discovered, as it frequently remains asymptomatic. Nevertheless, when symptoms manifest, the most common presentation involves recurrent pneumonia in a specific lung area. Additional symptoms may include persistent cough, back pain, or exertional dyspnea [6]. There have been limited cases documented in the literature wherein patients over the age of 50 have been diagnosed with PS [7–9]. In this report, we present an exceedingly rare case of intralobar sequestration in an elderly patient who has experienced recurring episodes of fever, chronic cough, and dyspnea over the past six years.

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2. Case presentation

A 62-year-old male patient, with a BMI of 28, presented with a chief complaint of recurrent episodes of fever, chronic cough, and difficulty breathing for the past six years. The patient denied experiencing palpitations, loss of appetite, weight loss, or hemoptysis. His medical history revealed a pattern of recurring pneumonia on the left side and angina. The pneumonia used to improve with antibiotic treatment. Regarding surgical history, the patient had previously undergone cardiac catheterization and stent placement. In terms of medication, he was taking daily doses of clopidogrel (75 mg), aspirin (81 mg), and atorvastatin (20 mg). He denied any history of smoking, alcohol consumption, or illicit drug use.

During the physical examination, auscultation of the lungs revealed decreased breath sounds and coarse crackles in the left infrascapular area. Other aspects of the physical examination were unremarkable. The patient's vital signs indicated a high body temperature of 38 °C and low oxygen saturation of 90%, while other vital signs were within normal ranges. Arterial blood gas analysis showed a low partial pressure of oxygen at 70%. Laboratory results, were within normal ranges. Echocardiography revealed mild mitral regurgitation with a normal ejection fraction of 50%. A chest computed tomography (CT) scan showed an abnormal development of lung tissue within the inferior lobe of the left lung. Notably, there was no identifiable connection between the anomalous tissue and the bronchial tree (Fig. 1). The anomalous tissue received arterial blood supply from the descending thoracic aorta above the diaphragm (Fig. 2). Venous drainage occurred through the pulmonary venous system to the left inferior pulmonary vein. These findings were indicative of an intralobar pulmonary sequestration.

Surgical intervention was deemed necessary. The patient underwent thoracotomy at the left sixth intercostal rib under general anesthesia. The anomalous artery originating from the descending aorta was ligated (Fig. 3). Subsequently, the anomalous tissue and artery were excised, and reconstruction of the left lower lobe was performed (Fig. 4). The incision was closed in layers, and a chest tube was inserted. The patient was then transferred to recovery without any complications. The chest tube was removed on the third day post-operation, and the patient was discharged on the fourth day without any significant events. A follow-up visit at the clinic two weeks after surgery indicated that the patient's health was satisfactory. However, subsequent regular contact could not be maintained.

3. Discussion

Pulmonary Sequestration (PS) comprises approximately 6% of the total prevalence of congenital pulmonary anomalies [10,11]. PS is a rare form of congenital pulmonary anomalies characterized by a portion of lung tissue that is non-functional and may be partially or entirely disconnected from the primary bronchopulmonary system. This anomalous lung tissue receives its blood supply from an aberrant branch originating from the aorta or its branches [12,13]. This finding is in accordance with our case, where the anomalous tissue was observed to receive arterial blood supply from the descending thoracic aorta positioned above the diaphragm. The

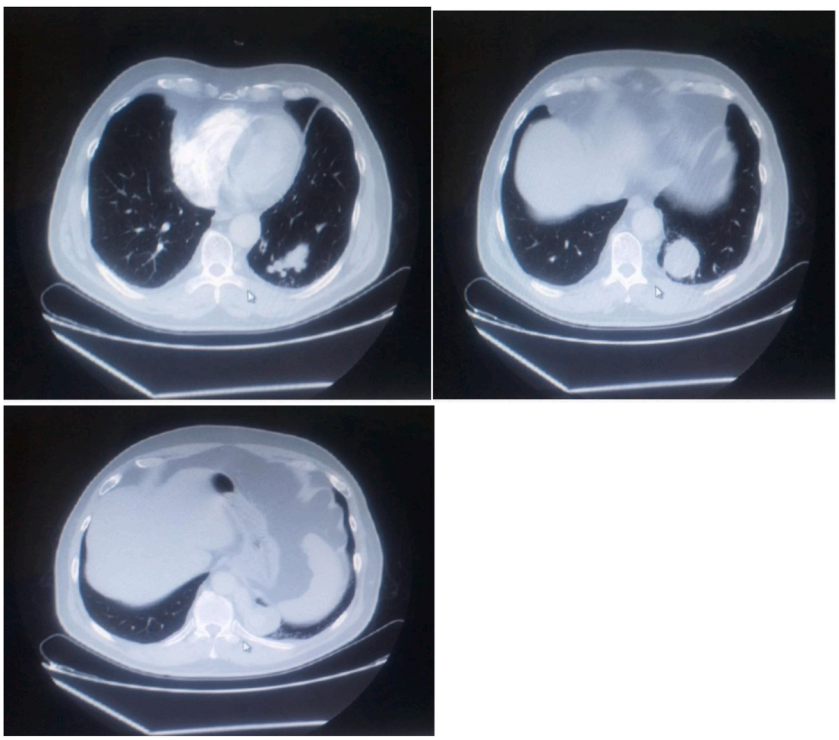

Fig. 1. A CT scan showing the anomalous segmental lung tissue in the left lower lobe.

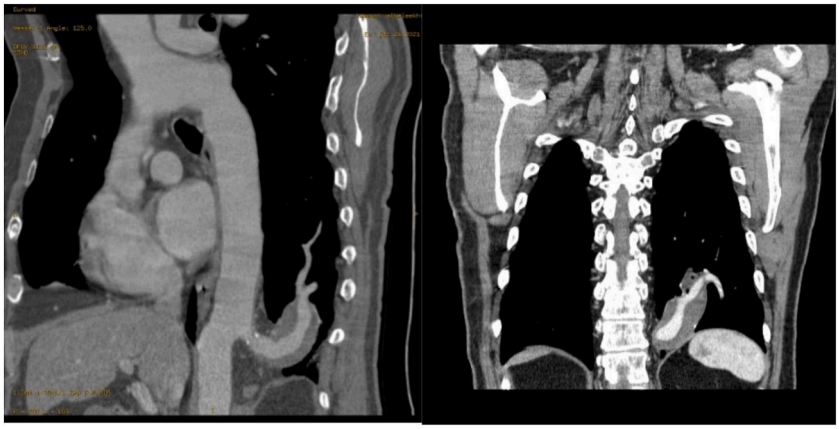

Fig. 2. Coronal CT views showing the arterial blood supply from the descending thoracic aorta above the diaphragm.

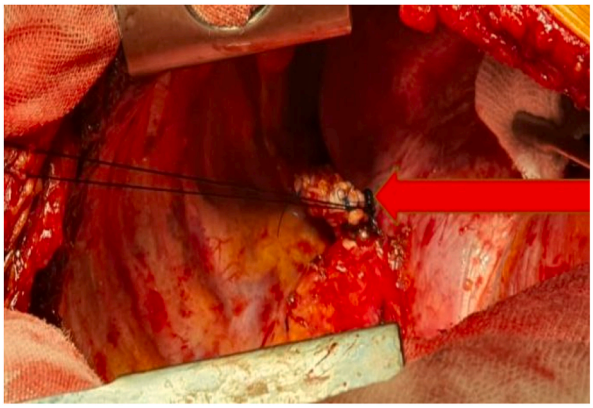

Fig. 3. Intra-operative photograph of the ligated anomalous artery arising from the descending thoracic aorta.

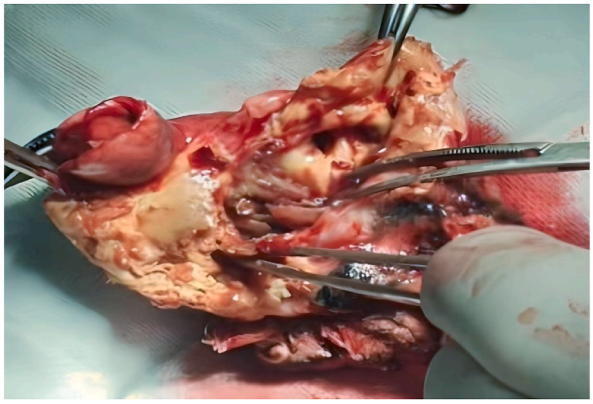

Fig. 4. Post-operative photograph of the resected anomalous tissue.

exact causes of pulmonary sequestrations are not fully understood, but various theories have been proposed to explain their origin [2]. One theory suggests that the primary abnormality lies in the development of primitive pulmonary tissue. This theory proposes that an accessory bud from the gastrointestinal tract develops into an accessory lung, which then acquires a systemic arterial supply. Another variation of this theory suggests that embryonic adhesions between the pulmonary bud and adjacent tissue separate the pulmonary tissue from the main anlage, leading to a sequestered tissue with a systemic arterial supply. However, Some authors have highlighted the importance of multiple factors such as infection, altered nutrition, heredity, and an imbalance of influences in a specific embryonic area that can lead to anomalies in the development of structures in that area [2].

PS can be categorized based on their anatomical positions as either intralobar sequestration (ILS), which is situated within the visceral pleura and encompassed by healthy lung tissue, or extralobar sequestration, which possesses an independent pleural covering

[14,15]. In reported cases, ILS stands out as the predominant form of PS, constituting a significant proportion of 75% [16]. The predominant site for the occurrence of ILS as in our case is observed within the posterior basal segments of the lung, with approximately two-thirds of cases manifesting in the left lung [17].

The clinical presentation of PS exhibits variability, contingent upon the specific characteristics encompassing the lesion, including its type, size, and location. The majority of cases typically lack overt symptoms, thus being predominantly asymptomatic [18]. Individuals presenting symptoms often display nonspecific features such as cough, fever, hemoptysis, and chest discomfort [19]. However, in our specific case, the elderly patient did not display the characteristic lack of symptoms observed in most reported cases. Instead, he presented with recurring episodes of fever, chronic cough, and respiratory difficulties.

Distinguishing PS from other conditions entails considering persistent pneumonia, lung abscess, congenital pulmonary airway malformation, bronchogenic cyst, pulmonary arteriovenous malformation, and Scimitar syndrome as differential diagnoses [20]. Prior to proceeding, it is essential to conduct either a CT scan of the chest or a Magnetic Resonance Imaging (MRI) to confirm the diagnosis, identify any anomalous arterial supply and venous drainage, and aid in the planning of surgical interventions [18]. The diagnosis of ILS typically occurs before the age of 20, manifesting through symptoms like recurrent pulmonary infections or cardiac disease. Conversely, it is uncommon for individuals to receive an ILS diagnosis after reaching the age of 40. In fact, there are only a few reported cases in the literature where patients over the age of 50 were initially diagnosed with this condition [7–9]. This serves to highlight the rare occurrence of our case, as it involves an elderly patient who was 62 years old, and that PS should be considered as one of the differential diagnosis even in elderly patients.

Surgical resection is the primary treatment modality recommended for PS to prevent recurring infections and potential hemorrhage. Although thoracoscopic resection has been associated with minimal morbidity and mortality, open thoracotomy was performed to our patient as it remains the recommended approach to effectively isolate and divide the abnormal systemic feeding arteries, thereby minimizing complications [21]. Even in asymptomatic patients, surgery is recommended to prevent further damage to the lung parenchyma. In situations where pulmonary infection or destruction of normal lung tissue occurs, more extensive resections such as lobectomy or pneumonectomy may be considered. Overall, surgical resection is highly successful and curative in the majority of PS cases [22,23].

4. Conclusion

In conclusion, Pulmonary Sequestration (PS) is a rare form of congenital pulmonary anomaly characterized by non-functional lung. The exact causes of PS remain unclear, but theories propose abnormalities in the development of primitive pulmonary tissue or embryonic adhesions. The clinical presentation of PS varies, with most cases being asymptomatic. While the diagnosis of ILS typically occurs before the age of 20, our case underscores the rare occurrence of PS in elderly patients, emphasizing the importance of considering it as a possible differential diagnosis. Surgical resection is the primary treatment modality, aiming to prevent complications and further lung damage. Open thoracotomy, as performed in our patient, is recommended to effectively isolate and divide abnormal feeding arteries. Future research should focus on understanding the underlying causes of PS, exploring the role of factors such as infection, altered nutrition, heredity, and embryonic influences.

Author statement

HH, AAN, AJ have participated in writing the manuscript. **HH, AAN, AJ** reviewed the literature. All Authors critically and linguistically revised the manuscript. **HH, AAN, AJ** contributed to revision of the manuscript. **HH** prepared and revised the final manuscript. **AS, KA, NA** supervised the conduct of the study and performed the surgery. All authors read and approved the final manuscript.

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Not applicable.

Availability of data and materials

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Consent for publication

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.

Declaration of competing interest

The authors declare that they have no competing interests.

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Not Applicable.

Abbreviation

(PS) Pulmonary Sequestration
(ILS) Intralobar Sequestration
(CT) Computed Tomography
(MRI) Magnetic Resonance Imaging

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