



► CASE REPORT

INTRALOBAR PULMONARY SEQUESTRATION AS A CAUSE OF RECURRENT PNEUMONIAS IN AN ADULT WOMAN: A CASE REPORT

Secuestro pulmonar intralobar como causa de neumonías recurrentes en una mujer adulta: reporte de caso.

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SUMMARY

Introduction: Intralobar pulmonary sequestration (IPS) is an uncommon congenital malformation in adults, characterized by non-functioning lung tissue supplied by an aberrant systemic artery. Its clinical and radiologic presentation may mimic recurrent pneumonias or pulmonary neoplasms, leading to diagnostic delays. **Case of report:** We report the case of a 49-year-old woman with a history of asthma and insulin resistance, who had recurrent right lower lobe pneumonias since 2018. In December 2023, during the most severe episode, initial chest CT suggested a possible pulmonary neoplasm; however, a second radiologic interpretation identified a basal pseudomass with an aberrant systemic artery consistent with pulmonary sequestration. Further imaging at referral centers confirmed anomalous arterial supply from the thoracic aorta. In May 2024, a right lower lobectomy was performed via thoracotomy, revealing an intraparenchymal aberrant artery. Pathology confirmed complicated IPS with fibrosis, bronchiectasis, and chronic inflammation. **Conclusion:** IPS should be considered in adults presenting with recurrent pneumonias and basal lung lesions. Clinical-radiologic correlation, particularly the identification of an aberrant systemic artery, is essential for establishing the correct diagnosis and guiding definitive treatment.

Keywords: Pulmonary sequestration; Pneumonia, Recurrent; Case Reports.
(Source: DeCS-BIREME)

RESUMEN

Introducción: El secuestro pulmonar intralobar (SPI) es una malformación congénita infrecuente en adultos, caracterizada por parénquima pulmonar no funcional irrigado por una arteria sistémica aberrante. Su presentación clínica puede simular neumonías recurrentes o masas pulmonares sospechosas de neoplasia, dificultando el diagnóstico oportuno. **Reporte de caso:** Presentamos el caso de una mujer de 49 años con antecedentes de asma y resistencia a la insulina, que desde 2018 experimentó episodios recurrentes de neumonía en el lóbulo inferior derecho. En diciembre de 2023, durante el episodio más severo, una tomografía inicial sugirió una posible neoplasia pulmonar; sin embargo, una segunda interpretación identificó una pseudomasa basal con una arteria sistémica aberrante compatible con secuestro pulmonar. Estudios en hospitales de referencia confirmaron irrigación anómala desde la aorta torácica. En mayo de 2024 se realizó una lobectomía inferior derecha mediante toracotomía, identificándose la arteria aberrante intraparenquimal. La anatomía patológica confirmó SPI complicado con fibrosis, bronquiectasias e inflamación crónica. **Conclusión:** el SPI debe considerarse en adultos con neumonías recurrentes y lesiones pulmonares basales. La correlación entre clínica e imagenología, especialmente la identificación de la arteria aberrante es fundamental para establecer el diagnóstico y definir el tratamiento adecuado.

Palabras clave: Secuestro pulmonar; Neumonía recurrente; Informes de casos.
(Fuente: DeCS-BIREME)

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► INTRODUCTION

Pulmonary sequestration is a rare congenital malformation, accounting for 1–6% of all reported pulmonary anomalies⁽¹⁾. It is characterized by non-functioning lung parenchyma that lacks communication with the bronchial tree and receives arterial supply from an aberrant systemic vessel^(2,3). Two variants have been described: intralobar pulmonary sequestration (ILS), the most common form, which shares visceral pleura with the normal lung; and extralobar sequestration (ELS), which is completely separated from the normal parenchyma and is more frequently diagnosed during childhood^(4,5).

ILS may present in adulthood, most often with recurrent pneumonias, hemoptysis, or basal pulmonary masses that mimic neoplasms, contributing to a high preoperative misdiagnosis rate reported to exceed 50%⁽⁶⁾. Radiologic similarity to primary lung tumors, complex bronchiectasis, or chronic infectious processes accounts for much of this diagnostic confusion. Additionally, elevated tumor markers such as CEA and NSE have been described in some cases^(7,8), which may further strengthen the erroneous suspicion of malignancy.

Diagnosis is primarily based on contrast-enhanced computed tomography with vascular reconstruction, which enables identification of the aberrant systemic artery—the pathognomonic hallmark of pulmonary sequestration^(1,9,10). Confirmation is usually complemented by angiographic studies or intraoperative findings. The treatment of choice is surgical resection through lobectomy, performed either via open thoracotomy or video-assisted thoracoscopic surgery (VATS), both of which offer favorable outcomes and low complication rates^(11,12).

This report describes the case of an adult woman with ILS initially interpreted as a pulmonary neoplasm, underscoring the diagnostic challenges and the importance of clinical–radiologic correlation to avoid misdiagnosis and guide appropriate management.

► CASE PRESENTATION

A 49-year-old woman, forensic physician and university professor, with a history of bronchial asthma since age 24,

insulin resistance, class II obesity, and chronic environmental exposure to pesticides due to residence near agricultural areas in Tumbes, presented with recurrent episodes of pneumonia in the right lower lobe beginning in 2018. These episodes were characterized by high fever, dry cough, and right-sided pleuritic chest pain.

First episode of pneumonia (August 2018). She presented with a fever of 39.5 °C, without prominent respiratory symptoms. Chest radiography revealed an image compatible with “atypical round pneumonia.” She was treated as an outpatient with azithromycin and acetaminophen, achieving early clinical resolution.

Second episode of pneumonia (June 2020). During the COVID-19 pandemic, she developed fever of 40 °C, dry cough, and general malaise. Non-contrast computed tomography revealed dilated and infected bronchiectasis in the right lower lobe. She was treated with intravenous ceftriaxone, metamizole, and acetaminophen, with progressive clinical improvement.

Third episode of pneumonia (December 2023). A fever of 40 °C, hyperoxia, and persistent cough prompted hospitalization for 15 days at Essalud. Laboratory tests showed leukocytosis of 19,000/mm³ with lymphocytosis. Intravenous ceftriaxone was initiated without clinical improvement, after which clindamycin was added. An initial contrast-enhanced chest CT scan reported a spiculated pulmonary nodule suggestive of neoplasia. A second interpretation performed at a private center described a medial basal pseudomass in the right lower lobe with bronchiectasis and an aberrant artery arising from the thoracic aorta, compatible with intralobar pulmonary sequestration (see Supplementary Figure 3).

Complementary studies. Additional CT scans performed at national referral hospitals between January and May 2024 confirmed a focal pseudonodular area in the right lower lobe, associated with an aberrant systemic vessel originating from the thoracic aorta (see Figures 1A, 1B, 1C).

Surgical intervention (May 2024). The patient was admitted to the Thoracic Surgery Department of the Guillermo Almenara Irigoyen National Hospital for definitive management. A right lower lobectomy was performed through a vertical axillary thoracotomy, entering via the fifth intercostal space.

An intraparenchymal aberrant systemic artery with branches arising from the right cardiophrenic sinus was identified. The inferior pulmonary vein, lower lobar artery, and lower lobar bronchus were ligated, and the surgical spe-

cimen was removed without intraoperative complications. (see Figure 2B)

Histopathological findings. The specimen revealed congested pleura, anthracotic lung parenchyma.



Figure 1. 1A. Chest CT angiography showing cystic bronchiectasis in the right lower lobe. 1B. Mediastinal window demonstrating the anomalous artery supplying the pulmonary sequestration (red arrow). 1C. Three-dimensional reconstruction of the thoracic aorta and its branches, with the red arrow indicating the origin of the anomalous systemic artery arising from the thoracic aorta, confirming pulmonary sequestration.

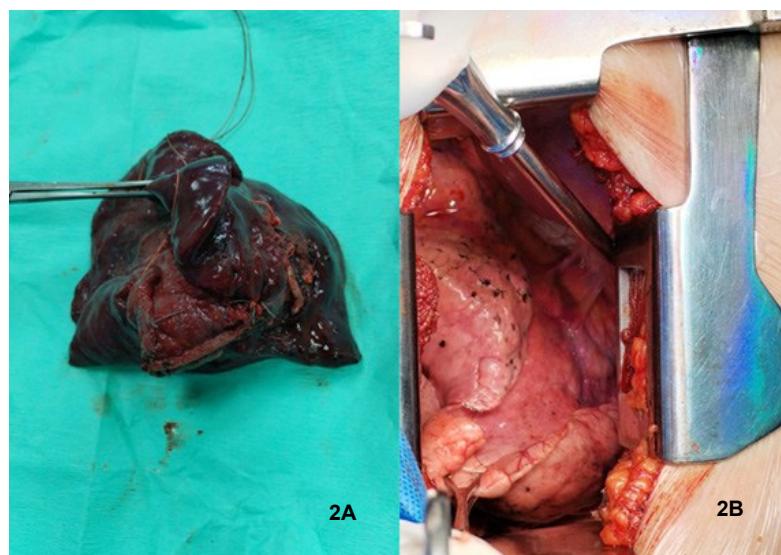


Figure 2. 2A. Resected surgical specimen showing discoloration of the right lower lobe and parenchymal fibrosis. Figure 2B. Aberrant arteries visualized arising from the thoracic aorta.

► DISCUSSION

Pulmonary sequestration (PS) accounts for approximately 1–6% of all congenital pulmonary malformations⁽¹⁾. Its incidence is estimated at 1 per 5,000 live births among all pulmonary pathologies⁽¹³⁾, though rates vary between 0.15–6.4% across different case reports and series^(14,15). In pediatric patients with congenital diaphragmatic hernia, an incidence of 3.4% has been described⁽¹⁶⁾.

Our case corresponds to intralobar pulmonary sequestration (ILS). A contrast-enhanced CT scan performed in December 2023 at Essalud reported a spiculated basal mass suspicious for lung neoplasm, without identifying any aberrant vascular structures⁽¹⁾. The absence of correlation with the patient's clinical history significantly limited the diagnostic accuracy of the initial radiologic assessment, ultimately leading to an incorrect diagnosis. In contrast, a second evaluation at an oncologic imaging center incorporated the patient's clinical context and successfully identified the aberrant systemic artery, the pathognomonic finding of ILS.

This illustrates a persistent diagnostic challenge: PS in adults is frequently misinterpreted as a neoplastic process. Several factors contribute to this issue, including: (a) atypical radiologic morphology involving multiple anomalous vascular structures and unconventional distribution patterns, which increase diagnostic complexity^(11,12); (b) pleural or basal localization; and (c) hypermetabolic uptake on PET/CT studies that mimics tumors, granulomatous infections, or chronic bronchiectasis^(2,3).

A retrospective series of 2,625 cases reported a preoperative misdiagnosis rate of 58.6%, with lesions frequently mistaken for pulmonary cysts (36.19%) or neoplasms (21.04%)⁽¹⁷⁾. Additionally, cases of ILS with elevated tumor markers such as carcinoembryonic antigen (CEA) and neuron-specific enolase (NSE) have been reported, further complicating the diagnostic process^(7,8). The presence of infection or hemothorax may also heighten the resemblance to malignant conditions⁽¹⁸⁾. To reduce diagnostic inaccuracies, multidisciplinary evaluation that integrates imaging and comprehensive clinical–radiologic analysis is recommended. Radiologists in both public and private centers typically have access to patients' clinical information and should incorporate it routinely.

ILS is more commonly diagnosed in pediatric and adolescent populations⁽¹⁹⁾, unlike the present case, which occurred after the age of 40. Therefore, ILS should be considered in the differential diagnosis of adults presenting with recurrent pneumonia. Unlike extralobar sequestration, ILS may drain into the bronchial tree and pulmonary veins, allowing partial ventilation and perfusion. This may delay symptom onset for decades⁽²⁰⁾. Furthermore, 60–70% of cases occur in the left lower lobe; however, in this patient, the lesion was located in the right lower lobe, a region where radiographic interpretation is often more challenging due to overlap with the cardiac silhouette⁽²¹⁾.

In Peru, a report by Nuñez-Paucar et al. at the National Children's Institute found that among 70 cases of congenital pulmonary malformations, 12% (9/70) were extralobar sequestration and 10% (7/70) were intralobar sequestration, in patients aged 0.5 to 168 months. Lobectomy was required in 87% of cases, with no associated mortality; postoperative pneumonia was the most frequent complication⁽²²⁾.

Lobectomy remains the most common and definitive surgical treatment for pulmonary sequestration. Conventional thoracotomy and video-assisted thoracoscopic surgery (VATS) are both associated with low complication rates and rapid recovery, particularly in cases of ILS^(4,5,11). Surgical resection prevents the recurrence of symptoms, infectious complications, and the exceedingly rare malignant transformation associated with sequestration⁽²³⁾. Segmentectomy or wedge resection may be considered in selected cases, but lobectomy remains the safest and most reliable option⁽¹²⁾.

► CONCLUSIONS

Integrating the patient's clinical history into radiologic interpretation is essential for accurately diagnosing rare entities. The identification of an aberrant systemic artery on contrast-enhanced imaging should raise a high index of suspicion for pulmonary sequestration. Surgical resection is curative and should be considered in symptomatic patients, particularly those with recurrent infections or persistent pulmonary lesions.

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