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CASE REPORT

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Pulmonary Sequestration Treated with Anatomical Segmentectomy: Case Report

Secuestro pulmonar tratado con segmentectomía anatómica: reporte de caso

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ABSTRACT

Pulmonary sequestration is a rare congenital lung malformation. It is characterized by an abnormal mass of dysplastic lung tissue supplied by an anomalous systemic artery separated from the normal bronchopulmonary tree. Misdiagnosis and inadequate treatment can lead to recurrent pneumonia and fatal hemoptysis. We present the case of a 43-year-old male patient diagnosed with pulmonary sequestration, accompanied by a brief review of clinical features, diagnostic approaches, and management options. Predominant symptoms were cough and hemoptysis. The contrast-enhanced chest computed tomography revealed an anomalous artery arising from the descending aorta, and it was decided to perform a resection of the left basal pyramid. Symptomatic patients with pulmonary sequestration should be treated surgically to avoid the risk of death from massive hemoptysis.

Key words: pulmonary sequestration; intralobar sequestration; hemoptysis; aberrant artery; surgery

RESUMEN

El secuestro pulmonar es una malformación pulmonar congénita poco común. Se caracteriza por una masa anormal de tejido pulmonar displásico irrigado por una arteria sistémica anómala y frecuentemente separada del árbol broncopulmonar normal. Un diagnóstico erróneo y un tratamiento inadecuado pueden provocar neumonías recurrentes y hemoptisis mortal. Presentamos el caso de un hombre de 43 años al que se le diagnosticó secuestro pulmonar y realizamos una breve revisión sobre las características clínicas, estrategias de diagnóstico y opciones de manejo. Los síntomas predominantes fueron tos y hemoptisis, la tomografía computarizada de tórax con contraste reveló una arteria anómala desde la aorta descendente, se decidió realizar resección de la pirámide basal izquierda. Los pacientes sintomáticos del secuestro pulmonar deben ser tratados mediante cirugía para evitar el riesgo de muerte por hemoptisis masiva.

Palabras clave: secuestro pulmonar, secuestro intralobar, hemoptisis, arteria aberrante, cirugía

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INTRODUCTION

Pulmonary sequestration (PS) is a relatively rare entity that accounts for 0.15% to 6.4% of all congenital pulmonary malformations. It generally consists of a systemic arterial supply to an abnormal pulmonary segment, associated with various forms of venous drainage.[1] In most cases, there is a single nutrient artery, although multiple systemic arteries may occasionally be present.[2] In the majority of cases, these segments do not have a connection with the tracheobronchial tree. Pulmonary sequestration is classified into two main types: intralobar and extralobar. The intralobar type is defined as a pulmonary segment contained within the same pleural lining as the adjacent lung, whereas the extralobar type has its own separate pleural covering.[1] Nearly 97% of cases are located in the lower lobe, with the left side being more common.[2] Sixty percent of these lesions are diagnosed within the first decade of life, and they are more common in males, with a 3:1 ratio.[1] Symptoms may vary depending on the type of sequestration. The extralobar form, which is often discovered through prenatal or neonatal ultrasound or magnetic resonance imaging (MRI), is frequently associated with other congenital anomalies, including cardiac defects, pulmonary hypoplasia, foregut duplication cysts, vertebral anomalies, or diaphragmatic anomalies. Infants may be asymptomatic or may develop respiratory distress due to pulmonary hypoplasia or a mass effect. Infection is rare because its pleural lining prevents contact with inhaled air. On the other hand, intralobar sequestration usually presents during childhood, although up to half of the cases appear after 20 years of age. Most patients remain asymptomatic for years, with diagnosis often prompted by a routine physical examination or after recurrent bacterial pneumonia in the affected lobe. Symptoms are usually nonspecific; the most common includes cough or sputum production, fever, hemoptysis, and chest pain.[2] Sequestration may be incidentally discovered in radiographic studies. Confirmation requires computed tomography angiography (CTA) or magnetic resonance angiography (MRA), which show the systemic feeding artery and its venous drainage.[3] The arterial supply is variable: 74% originates from the thoracic aorta, while the remainder arises from the abdominal aorta and its branches, including the gastric or splenic arteries. Venous drainage from these pulmonary segments typically occurs through the pulmonary venous system, although systemic venous drainage has also been reported.[1]

CASE REPORT

A 43-year-old male patient, previously healthy and a swimmer, was admitted due to frank hemoptysis associated with episodes of nonproductive cough, with no other accompanying symptoms.

Physical examination revealed a healthy-appearing patient. Vital signs, including oxygen saturation on room air, were within normal limits. Blood residues were observed on the mucosa of the palatal vault.

Laboratory tests –including complete blood count, renal function tests, electrolytes, D-dimer, and inflammatory markers–were negative. The initial chest computed tomography (CT) revealed atelectatic changes in the left lung base, involving the anterior and lateral segments, superimposed with ground-glass opacity and airway lumen filled with secretions. In addition, an anomalous origin of the left lower lobar artery arising directly from the descending aorta was observed, with venous drainage directly into the right atrium, and no evident abnormality of the tracheobronchial tree.

Hospitalization in a closed unit was indicated for clinical monitoring. A computed tomography angiography (CTA) with 3D reconstruction was performed, which demonstrated pulmonary parenchyma of homogeneous density with a heterogeneous opacity containing areas of ground-glass attenuation and others with a consolidative appearance, involving the entire basal pyramid of the left lower lobe. There was also compensatory hyperinflation of the apical segment of the left lower lobe. The mediastinum showed normal morphological features, with a systemic artery arising from the distal thoracic aorta that courses through the triangular ligament of the mediastinum toward the left lower lobe. The venous drainage and bronchial tree were normal. **(Figure 1)**

The patient remained hospitalized for four days in stable condition. During this time, he experienced one episode of coughing with hemoptoic sputum, without hemodynamic alterations or laboratory changes.

Considering the clinical history of the patient, the imaging findings compatible with intralobar sequestration, and the persistence of symptoms, resection of the left basal pyramid was indicated.

A left video-assisted thoracoscopic surgery (VATS) was performed, identifying the anomalous systemic artery supplying the basal pyramid. The artery was repaired and subsequently ligated and sectioned. Then, a basal pyramid segmentectomy was carried out following the standard surgical technique. **(Figure 2)**

DISCUSSION

Intralobar pulmonary sequestration is a relatively rare congenital anomaly, typically diagnosed in childhood but occasionally identified in adulthood.[2] Intralobar sequestration (ILS) is more common than extralobar sequestration (ELS), accounting for about 75% of cases. Patients may present with an incidental pulmonary lesion detected on imag-

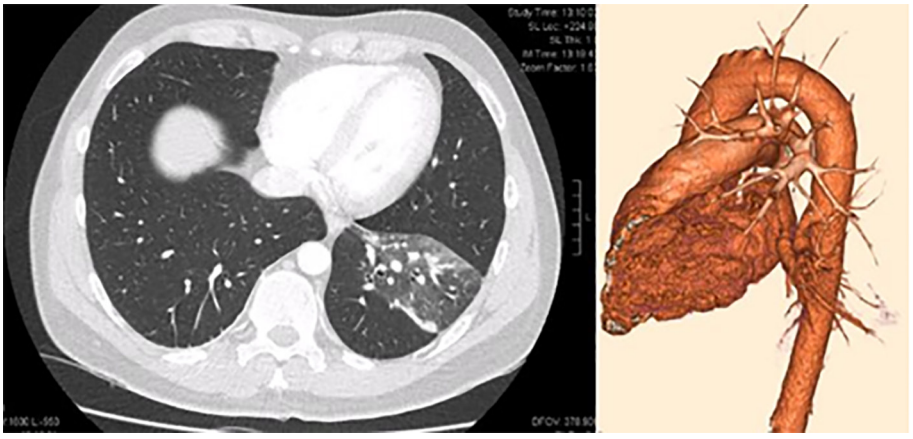

Figure 1. CT angiography: heterogeneous opacity in the basal pyramid of the left lower lobe. 3D reconstruction: evidence of an anomalous artery arising from the thoracic aorta.

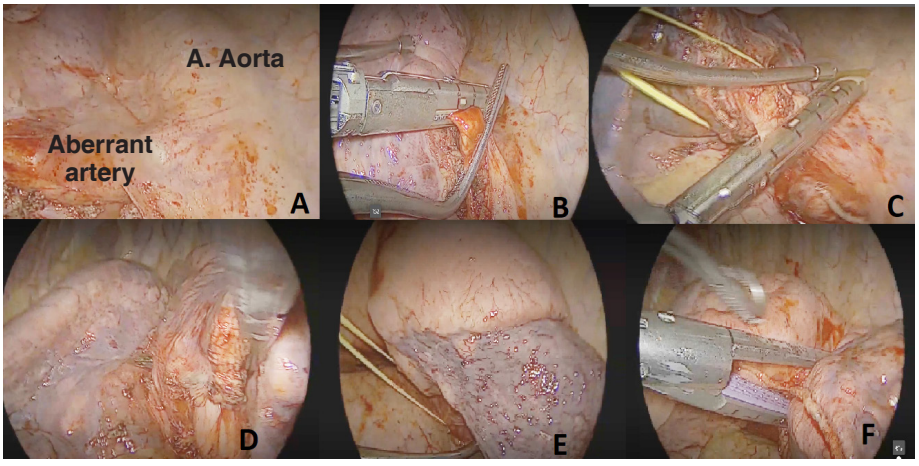

Figure 2. Steps of the anatomical segmentectomy A) Video-assisted thoracoscopic surgery showing the anomalous systemic artery. B) Repair and sectioning of the anomalous artery. C) Identification and sectioning of the vein of the basal pyramid. D) Identification and sectioning of the artery of the basal pyramid. E) Clamping of the bronchus of the basal pyramid and collapse of the segment. F) Sectioning of the pulmonary parenchyma corresponding to the basal pyramid.

ing and otherwise remain asymptomatic. However, the most common presentation includes recurrent pneumonia, chronic cough, and hemoptysis. Few reports have described more severe complications, such as superimposed aspergillosis or even fatal hemoptysis.[4] For these reasons, sequestration has been traditionally treated with definitive resection of the affected pulmonary segment.

In our experience, contrast-enhanced chest computed tomography enabled diagnosis and delineated key anatomical features for surgical planning.

Definitive treatment involves resection of the affected pulmonary segment. Several key elements must be considered, including preoperative antibiotics for pneumonia exacerbation and precise preoperative identification of arterial supply.[1] The extent of resection aims to preserve as much normal lung tissue as possible, justifying the anatomical segmentectomy when feasible. Lobectomy is appropriate when sequestered tissue is difficult to distinguish from functional parenchyma.[3]

An alternative approach must be mentioned: exclusion of the aberrant arterial supply via endovascular embolization using several occlusion devices, indicated in neonates with large-caliber pulmonary sequestration and severe hemodynamic instability.[5] However, embolization raises main concerns, including possible incomplete vascular occlusion, subsequent evolution of sequestered tissue, and symptom recurrence.[2]

In our experience, surgical resection remains the preferred approach in symptomatic patients due to the risk of potentially fatal hemoptysis and infectious recurrence.

Conflict of interest
Authors have no conflicts of interest to declare.

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