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Chest

Case report: Pulmonary sequestration in an adult

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ABSTRACT

Pulmonary sequestration is a rare congenital malformation that is uncommonly diagnosed during adulthood. Pulmonary sequestrations can manifest with variable presentations. It can remain asymptomatic or present with more severe symptoms such as hemoptysis and recurrent pneumonia. Diagnosis can be confirmed with computed tomography angiogram or angiography. Treatment with embolization of the afferent artery and surgical removal of the pulmonary sequestration generally has good outcomes. We report a case of a 43-year-old man who presented with recurrent pneumonia and left-sided back pain and was eventually diagnosed with pulmonary sequestration.

Introduction

Pulmonary sequestration is a rare congenital lung anomaly defined as an area of dysplastic and nonfunctioning pulmonary tissue with an anomalous systemic blood supply and various forms of venous drainage [1]. The incidence of congenital pulmonary airway malformations is 1 per 8300 to 35,000, and pulmonary sequestrations compromise 0.15% to 6.4% of congenital lung malformations [2]. Pulmonary sequestration is primarily considered a childhood disease, as most cases are diagnosed early in life. However, in some cases, the diagnosis may not be evident during childhood and individuals with this condition can continue their lives experiencing mild or no symptoms. Asymptomatic pulmonary sequestrations are typically found incidentally on diagnostic radiology imaging. Pulmonary sequestrations most commonly present as recurrent pneumonias; however, it has also been reported to present with chest pain, cough, shortness of breath, and hemoptysis [3]. We report a case of a 43-year-old man who presented with recurrent pneumonia and left-sided back pain and was eventually diagnosed with bronchopulmonary sequestration.

Case report

A 43-year-old Hispanic man presented with the chief complaint of left-sided back pain for 2 weeks. The pain gradually increased in severity since he completed a full course of antibiotics for an unresolving pneumonia that started 1 month ago. The patient admitted to having a cough and mild fever. He denied chest pain, shortness of breath, hemoptysis, and weight loss. His medical history was significant for recurrent

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pneumonia during childhood. Surgical history was significant for appendectomy during childhood and a gastric bypass 10 months before admission. He denied smoking, alcohol, and illicit drug use. On physical examination, all his systems were unremarkable. His vital signs were within normal limits.

Laboratory results including complete blood count, complete metabolic panel, and coagulation studies were unremarkable. Chest x-ray revealed a left basilar parenchymal opacity that was initially concerning for malignancy. A follow-up computed tomography (CT) scan showed that the lesion was a large mass in the left lower lobe measuring 10.4×7.8 cm in axial dimensions. Additional laboratory studies of CEA, quantitative beta-HCG, and AFP tumor markers were negative.

A CT-guided biopsy was performed because of concern of a possible malignancy. Pathology determined that the biopsy was negative for any atypical or malignant cells and contained bronchial epithelial cells, macrophages, and other inflammatory cells.

During further review of the CT scan, an arterial vessel that arose directly from the posterior lateral aspect of the aorta supplied the left lower lobe mass, consistent with a pulmonary sequestration.

Interventional radiology performed an aortic arteriogram. This demonstrated a systemic artery arising from the distal descending thoracic aorta supplying the left lower lobe sequestration with numerous abnormal corkscrew-type arteries. There was also abnormal venous drainage from the sequestration into the coronary sinus and right atrium. Interventional radiology then performed ultrasound and fluoroscopic-guided coil embolization of the systemic arterial feeder to the pulmonary sequestration.

The patient eventually underwent a left thoracotomy with resection of the left pulmonary sequestration. Grossly, the partial lobe of lung measured $13 \times 10 \times 5$ cm and weighed 320 g. The lung tissue exhibited dilated air spaces with exudate, organizing pneumonia, and thick arteries, consistent with the clinical diagnosis of pulmonary sequestration.

After surgery, the patient was stabilized and discharged home without complications.

The patient was lost to follow-up, and unable to sign consent after multiple attempts to contact the patient. All personal identifiable information has been removed in this article.

Discussion

A pulmonary sequestration is a very rare anomaly that can present in adults. The aberrant anomaly represents a segment of pulmonary tissue without connection to the tracheobronchial tree or pulmonary arterial circulation [4]. Pulmonary sequestrations are categorized based on their anatomical locations, as either intralobar or extralobar [5].

Extralobar sequestrations are covered by a layer of the visceral pleura and are completely separated from the functioning areas of lung; in contrast, intralobar sequestrations are not separated by a layer of pleura and are located within normal lung tissue [5]. In this case, the patient had an intralobar sequestration, which in 60% of patients is diagnosed at age 20 or younger and is rarely found in adults older than 40 years [6].

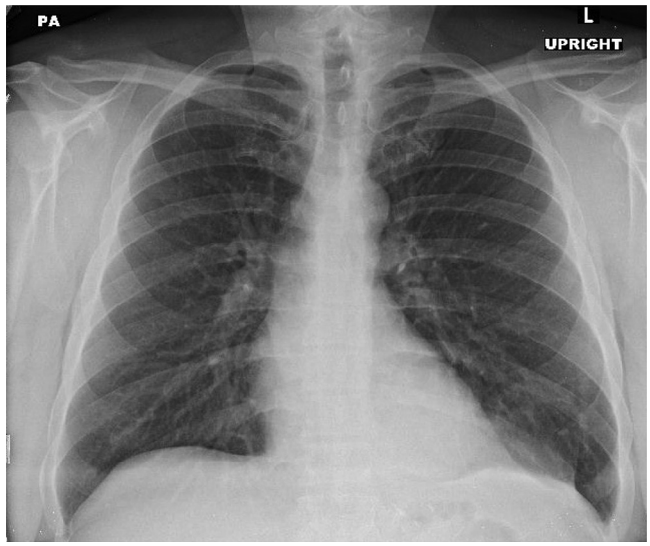

Fig. 1 – Chest radiograph demonstrates an opacity in the left lower lobe.

Most pulmonary sequestrations arise in the left lung. Approximately two-thirds of pulmonary sequestrations are found in the posterior basal segment of the left lower lobe [4]. They receive blood supply from the systemic circulation, most commonly the thoracic or abdominal aorta [5]. Pulmonary sequestrations have venous drainage into either the pulmonary veins, azygous vein, hemiazygos vein, inferior vena cava, or right atrium [5].

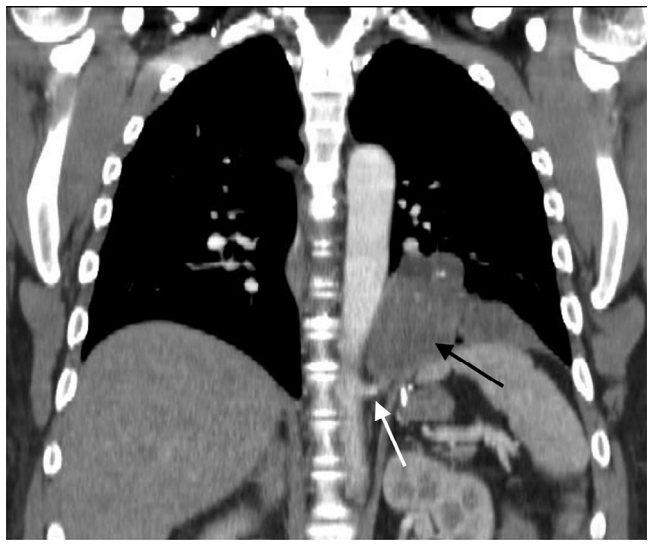

Fig. 2 – Chest computed tomography with intravenous contrast showing a section of the pulmonary sequestration (black arrow) with the systemic feeder artery from aorta (white arrow).

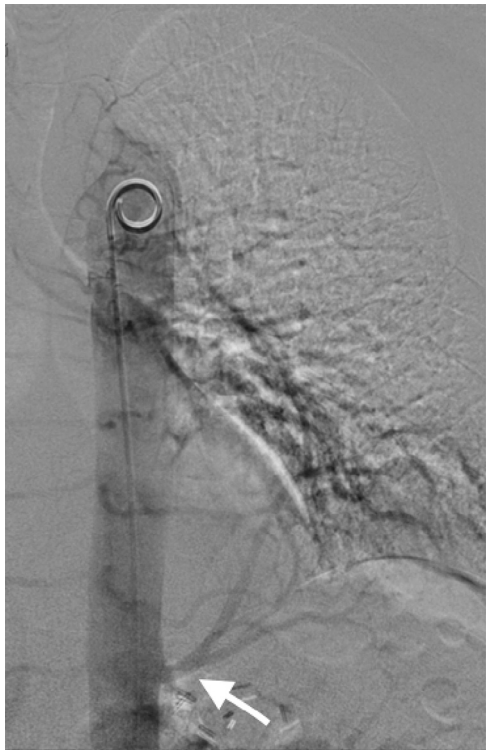

Fig. 3 – Aortic arch arteriogram, showing a systemic arterial feeder to the pulmonary sequestration in the left lower lobe.

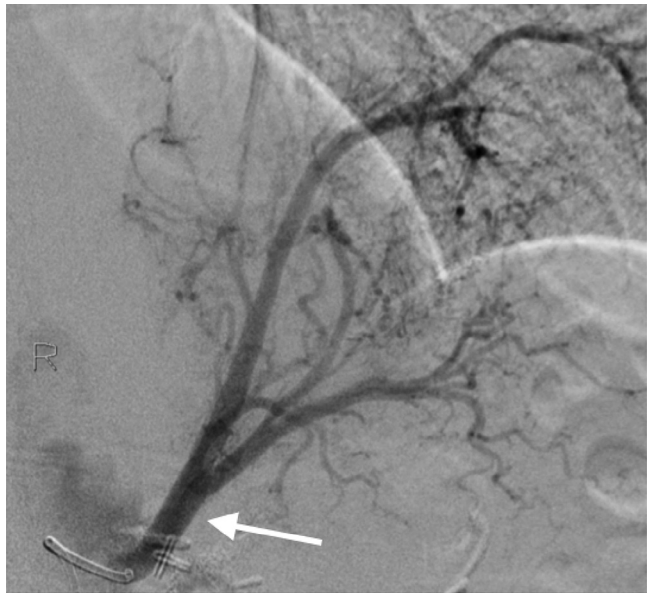

Fig. 4 – Digital subtraction angiography demonstrating a branch from the aorta supplying the pulmonary sequestration in the left lower lobe.

Microscopically, intralobar sequestrations are characterized by cystic lesions, and the lung parenchyma displays inflammatory tissue and fibrosis along with lymphocyte-rich infiltrated tissue [5]. These cysts are typically lined by cuboidal or columnar epithelium; additionally, alveoli involved in the sequestration may display characteristics of hyperinflation and emphysema-like changes, along with bronchiectasis [5].

The diagnosis of pulmonary sequestrations can be accomplished through CT angiography [1,7]. CT angiography offers a noninvasive approach compared with catheter aortography, which used to be necessary for determining the arterial supply and diagnosis [1]. However, in our case, the afferent vessel feeding the pulmonary sequestration was visualized through a postcontrast CT not specifically performed as CT angiography.

The optimal treatment for a pulmonary sequestration involves surgical resection to avoid infection and damage to the lung parenchyma [7]. Surgery is recommended even in asymptomatic patients [1,7]. In cases of pulmonary infection or destruction of normal lung tissue, a major resection such as lobectomy or pneumonectomy may be considered [7]. The outcome is usually curative (Figs. 1-4).

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