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Case Report

Embolization of symptomatic intralobar pulmonary sequestration – A minimally invasive treatment option

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ABSTRACT

The growing field of interventional radiology provides a number of treatment options alternative to previous relied upon invasive surgeries. Pulmonary sequestration is a condition where the option of interventional radiology is particularly promising. Described is a 41-year-old male who elected alternative treatment for his symptomatic intralobar sequestration in order to avoid associated complications of extensive surgery. Specifically, this case reports a successful cure utilizing minimally invasive embolization with polyvinyl alcohol particles and coil placement.

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Introduction

Pulmonary sequestration is a rare condition only representing 0.15%–6.4% of congenital lung malformations [1]. It is described as nonfunctioning dysplastic lung tissue resulting in a variety of pulmonary symptoms depending on the tissue's relation to the lung parenchyma. Surgical resection is the standard of care for both intralobar and extralobar pulmonary sequestration.

Case report

A 41-year-old male smoker with well-controlled hypertension presented with recent onset of recurrent pneumonias. He denied a history of pneumonia as a child or adolescent. A chest CT was performed demonstrating a mass in the postero-medial left lung base with a large feeding artery originating directly from the distal thoracic aorta and prominent venous return to the left lower pulmonary vein, most consistent with an intralobar pulmonary sequestration (Fig. 1). The patient was evaluated by cardiothoracic surgery for resection

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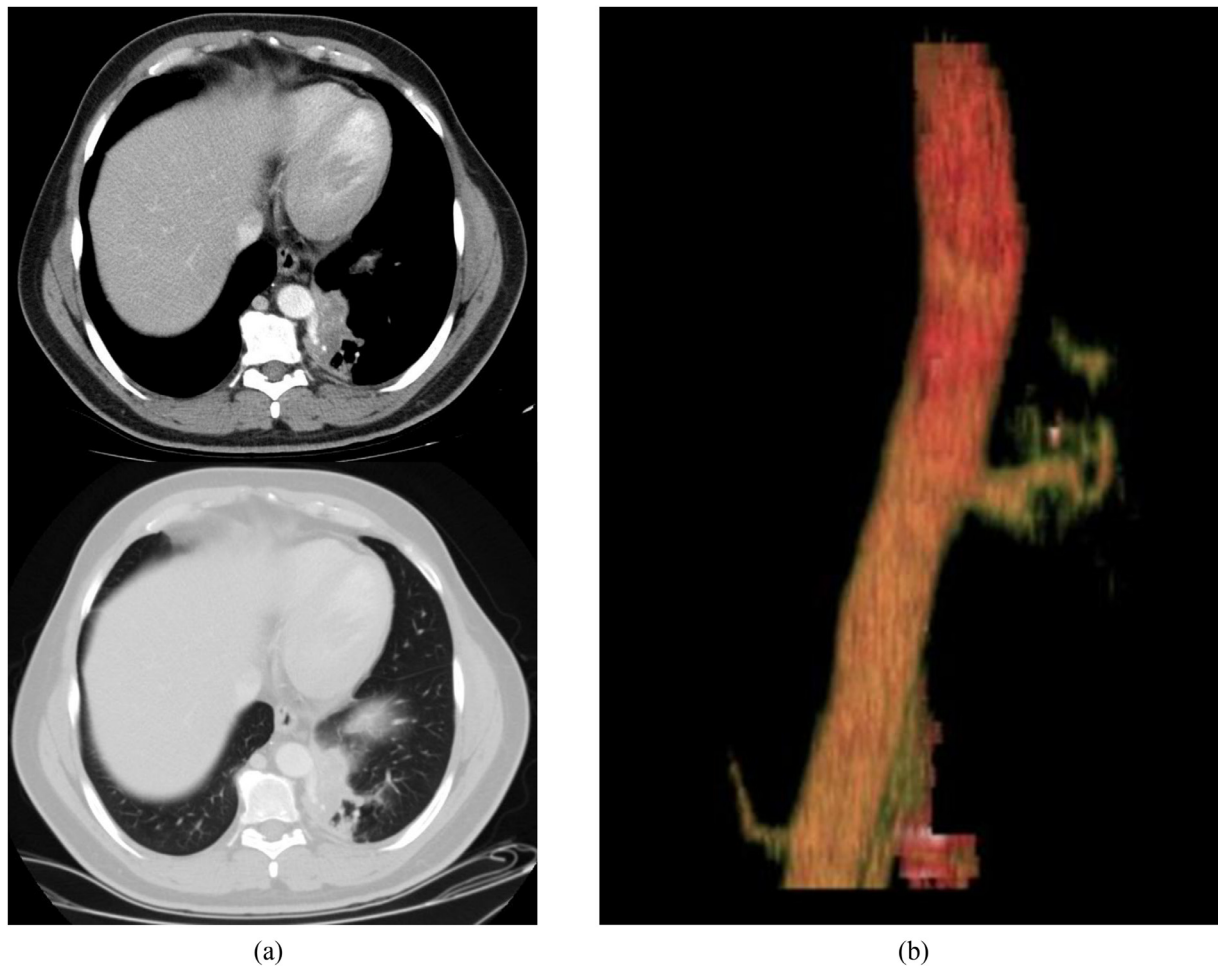


Fig. 1 – (a)-Axial chest CT with contrast demonstrating a consolidation in the posteromedial left lower lobe with a large feeding artery originating directly from the thoracic aorta. (b)-3D reconstruction of the descending aorta showing the feeding artery from the aorta.

of the sequestration. However, after extensive discussion of the risks and benefits the patient desired to pursue a less invasive treatment method. He was referred to interventional radiology to discuss embolization as a treatment option, and decided to pursue this as a minimally invasive alternative to treat his sequestration. After confirming normal baseline spirometry and no residual evidence of pneumonia, the patient was scheduled with interventional radiology to undergo diagnostic angiography with therapeutic embolization in order to avoid surgery and its potential complications.

Access was gained through the right common femoral artery with a 5F micropuncture kit using standard technique. A vascular sheath was then advanced into the aorta and several digital subtraction arteriograms were performed to identify and delineate the sequestration's feeding artery. A 0.016 Fathom wire (Boston Scientific, Marlborough, MA) and Renegade Hi-Flo microcatheter (Boston Scientific, Marlborough, MA) were then advanced into the dominant artery supplying the sequestration (Fig. 2). More distal feeding

arteries of the lesion were subselected and 1000 micron PVA particles (Cook Medical, Bloomington, IN) were slowly injected until near complete stasis of blood flow was observed. After embolizing with PVA particles, the vessels were coiled utilizing multiple platinum Cook Tornado and Nester coils (Cook Medical, Bloomington, IN) of varying sizes (Fig. 3). This method was repeated to occlude all arterial branches supplying the sequestration. Four Boston Scientific Interlock coils were also used to occlude a central aneurysmal portion of the sequestration. At the conclusion of the procedure, complete stasis was confirmed with arteriography (Fig. 4).

The patient tolerated the procedure well and there were no complications. He experienced some pain which was controlled with nonsteroidal anti-inflammatory drug and completely resolved by 10 days after the procedure. A follow-up CT redemonstrated multiple coils leading up to and within the sequestration with no evidence of new collateralization or infection. The patient continued to do well and remained pneumonia free.

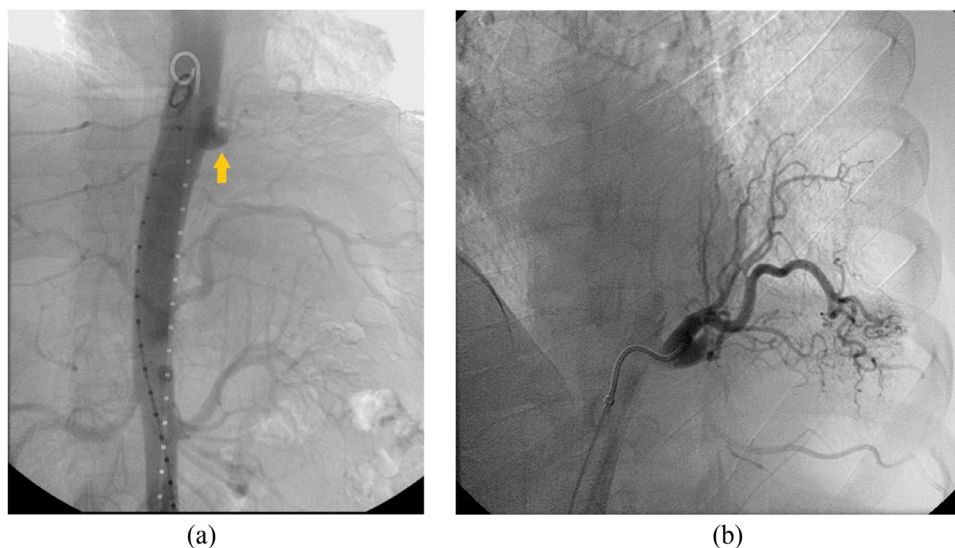


Fig. 2 – (a)-Digital subtraction angiogram (DSA) of the aorta which contains a flush catheter and identifies of the origin of the sequestration feeding artery (orange arrow). **(b)**-DSA showing the extensive arterial supply to the left lower lobe sequestration. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article).

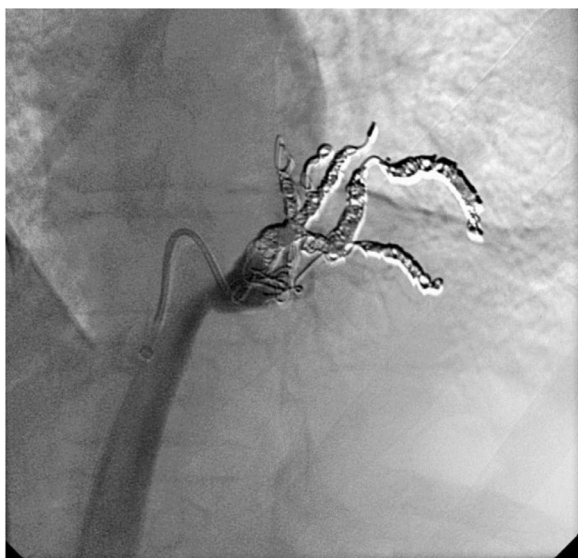


Fig. 3 – DSA after the deployment of coils throughout the arterial supply of the sequestration.



Fig. 4 – DSA of the aorta demonstrating successful embolization of the left lower lobe pulmonary sequestration with complete arterial stasis.

Discussion

Pulmonary sequestration is a congenital anomaly resulting in nonfunctioning pulmonary tissue with no communication to the tracheobronchial tree and systemic blood supply. The condition is classified as either intralobar, inside the lung without its own pleura, or extralobar, outside the lung with its own pleura, each presenting differently. Intralobar sequestrations are often asymptomatic until adulthood and manifest as recurrent pneumonias, shortness of breath, cough, and

sometimes hemoptysis. Extralobar sequestration has a worse prognosis and can present in infancy with respiratory distress, heart failure secondary to a right to left shunt, and pulmonary hemorrhage [2]. Intralobar sequestration identification is more straightforward than extralobar. Definitive diagnosis is accomplished by confirming systemic arterial blood supply with venous drainage specific to the sequestered lung tissue. Imaging is used to demonstrate this abnormal arterial supply and to exclude other thoracic pathologies [3]. The dominant artery supplying the sequestration often originates

from the thoracic aorta, abdominal aorta, celiac trunk, or left gastric artery. Other common radiologic manifestations include mass/consolidation, hyperlucency, cystic changes, and dilated bronchi [4]. Extralobar sequestration can be difficult to differentiate from retroperitoneal tumors and requires a more involved evaluation including catecholamines, cortisol, aldosterone, and tumor markers.

Surgical excision is the current standard of care and can be achieved with either open thoracotomy or video-assisted thoracoscopic procedures for symptomatic patients. Treatment of asymptomatic patients is more controversial, but still recommended due to potential future complications. Symptomatic intralobar sequestration specifically, as in our patient's case, often requires lobectomy. Recurrent infection secondary to the intralobar sequestration results in friable vasculature and increased risk for intraoperative hemorrhage [5]. Additionally, a retrospective review from Mayo Clinic reported a 28% postprocedure complication rate describing bronchopulmonary fistula resulting in air leak, hemoptysis, empyema, and arrhythmia [4]. Endovascular embolization and coiling is a recently established alternative therapy being utilized more frequently in order to avoid these complications. Embolization occludes the arterial supply resulting in sequestration regression by infarction, necrosis, and fibrosis [5]. Embolic agents include polyvinyl alcohol particles, micro coils, n-butyl cyanoacrylate glue, Amplatzer occlusion devices, alcohol, and gelatin [6]. General complications of embolization include fever, access site thrombosis, transient limb ischemia, and nontarget embolization [5]. Sequestration may also recur status postembolization due to formation

of shunts, incomplete closure, and displacement of embolic agents. This patient represents one of the limited reported cases of curative embolization with no long-term complications. Further research about the long-term outcomes of those patients who undergo sequestration embolization is necessary for better evaluation of this promising minimally invasive alternative treatment option.

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