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Case Report

Interventional management of aspergillus-related pulmonary artery pseudoaneurysm and hemoptysis *

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ABSTRACT

Pulmonary artery pseudoaneurysms are rare but life-threatening vascular abnormalities. Only less than 10% hemoptysis cases are of pulmonary artery origin while most cases arise from bronchial arteries. When diagnosed, they are mostly found to accompany pre-existing cardiovascular disease, infection, (i.e. Tuberculosis or Aspergillosis), vasculitis, trauma and/or neoplastic conditions. There are rare reports of pulmonary artery pseudoaneurysms being caused by direct extension of invasive fungal infections. We report a case of a rapidly growing pulmonary artery pseudoaneurysm in a 20-year-old female with lymphoma involving the lung and mediastinum. The patient was hospitalized with complications, including hemoptysis in the setting of Aspergillus Pneumonia and respiratory failure requiring intubation. Interventional Radiology was consulted after multiple bronchoscopic interventions failed to stabilize the bleeding. Patient then underwent embolization of the left subsegmental pulmonary artery pseudoaneurysm, with resolution of hemoptysis the next day.

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Introduction

Hemoptysis remains a common cause of consultation for Interventional Radiology in an emergency setting. The reported mortality rate from hemoptysis ranges from 9% to 38% and

depends on bleeding rate and its etiology [1]. In massive hemoptysis, airway maintenance is of first and paramount importance due to risk of asphyxiation [2]. Bronchoscopy remains an important first line therapy in respiratory decompensation due to hemoptysis. If bronchoscopy does not yield stabilization and the source is not identified, then further di-

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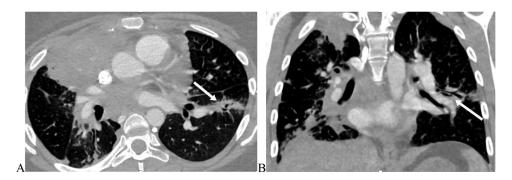


Fig. 1 – (A,B) Contrast enhanced computer tomographic axial (A) and coronal (B) views of the thorax 10 weeks prior to diagnosis of pseudoaneurysm demonstrate left lower lobe consolidation (white arrows) without pseudoaneurysm.

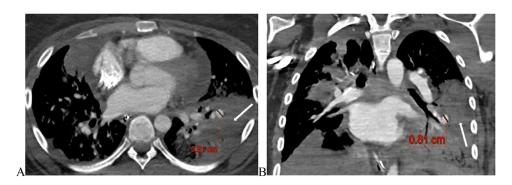


Fig. 2 – (A,B) Contrast enhanced computer tomographic axial (A) and coronal (B) views of the thorax show left lower lobe pulmonary artery pseudoaneurysm measuring 0.8 cm approximately 4 weeks prior to intervention. White arrows show depict the consolidation surrounding the pseudoaneurysm.

agnostic imaging can be obtained. Bronchial artery is estimated to be the source of bleeding in \sim 90% of massive hemoptysis, while systemic collateral vasculature, such as internal mammary or intercostal arteries account for another 5% of hemoptysis [1]. We describe a rare case of hemoptysis related to pulmonary artery pseudoaneurysm, which in the literature is estimated to be the cause of hemoptysis in only 5% of patients [1].

Pulmonary artery pseudoaneurysm is defined as a contained rupture of the pulmonary artery [3]. As such, a timely diagnosis requires clinicians to be aware of its imaging features and continue to be suspicious of its possibility [3]. When they are diagnosed, they are mostly found to be acquired in setting of pre-existing cardiovascular disease, infection, vasculitis, trauma and/or neoplastic conditions [3]. Now, with more safe and efficacious endovascular techniques, surgical intervention of pulmonary artery pseudoaneurysms has been widely replaced by embolization [1].

Case report

A 20-year-old female with progressive diffuse large B-cell lymphoma despite multiple lines of therapy presented to urgent

care for hemoptysis. On presentation, patient reported a cough with maroon colored sputum for 1 day. She also reported pain on the right side of her chest, fatigue, low grade fevers, chills, and exertional dyspnea. On initial evaluation, patient was on room air with episodes of small volume hemoptysis. Patient's prior CT from 10 weeks ago was notable for pulmonary and mediastinal masses related to history of lymphoma and no vascular abnormality (Figs. 1A and B). She was admitted for conservative care and further imaging workup. Follow up CT angiogram of the chest showed an 8.0 mm pulmonary artery pseudoaneurysm in the left lower lobe (Figs. 2A and B). Blood cultures returned positive for grampositive bacteremia. She was then managed conservatively with antibiotics and respiratory support as needed. After improvement, she was discharged home with anticipation for initiation of CAR-T treatment.

Shortly after her discharge, the patient presented back for recurrent hemoptysis and was re-admitted for management of hemoptysis and pneumonia. CT angiogram of the chest showed an increasing in size left lower lobe pulmonary artery pseudoaneurysm, now measuring 1.2 cm with surrounding consolidation (Figs. 3A and B). The patient underwent bronchoscopy with bronchoalveolar lavage revealing left lower lobe blood clot with segmental distribution (anteromedial) matching that of the location of the PSA. The patient was

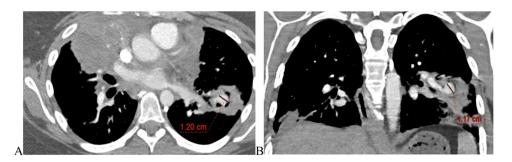


Fig. 3 – (A,B) Contrast enhanced computer tomographic axial (A) and coronal (B) views of the thorax show interval enlargement of the pseudoaneurysm measuring 1.2 cm.



Fig. 4 – Selective digital subtraction angiography of the left lower lobar pulmonary artery branch demonstrating the pseudoaneurysm (red circle).

then referred to Interventional Radiology for management of recurrent hemoptysis. She was then evaluated for an urgent pulmonary angiogram with embolization with general anesthesia due to patient's ASA status (IV-V) and need for airway protection.

In IR, the right common femoral vein was accessed, and pulmonary artery was cannulated with 6 French angled pigtail catheter. After angiography of the main pulmonary artery, a 5 French angled diagnostic catheter was used to access the left lower lobe PA (Fig. 4). Selective angiography of a segmental pulmonary branch was then performed, and the origin of the pseudoaneurysm was identified. A 2.8 French microcatheter was then used to select the segmental branch supplying the pseudoaneurysm and the microcatheter was advanced into the pseudoaneurysm lumen for embolization. A combi-

nation of fibered pushable and detachable coils were used. Care was taken to anchor the detachable coil at an adjacent small branch for enhanced stability (Fig. 5). Post embolization angiography demonstrated complete exclusion of the pseudoaneurysm.

On follow up, the patient reported coughing a small amount of "old clot" the next morning following the procedure with subsequent resolution of hemoptysis. Follow up CT thorax within 2 weeks demonstrated stable coil pack with adjacent small parenchymal changes and no evidence of recanalization (Fig. 6). At this time, left lower lobe bronchoalveolar lavage fungal culture returned with Aspergillus, the most likely etiology for development and rapid progression of pulmonary artery pseudoaneurysm. The patient was then started on voriconazole.

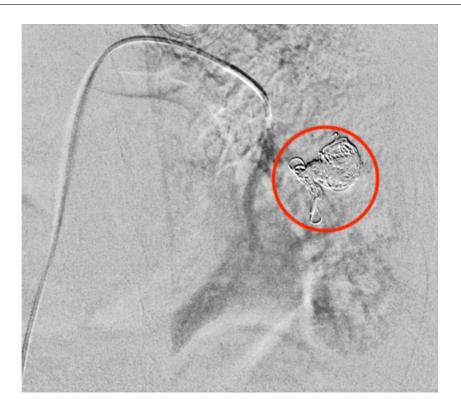


Fig. 5 – Digital subtracted angiography after coil embolization (red circle) demonstrates complete exclusion of the pseudoaneurysm with no antegrade flow.



Fig. 6 – Follow up contrast enhanced computer tomography of the thorax shows coil pack (white arrow) at the site of the pseudoaneurysm with no cavity filling consistent with successful treatment.

Discussions

Pulmonary artery pseudoaneurysms are very rare and can arise secondary to infection, malignancy, congenital heart disease, and/or vasculitis. We describe a case of an immunocompromised patient who developed Aspergillus pneumonia which eventually resulted in symptomatic pulmonary artery pseudoaneurysm.

Both true aneurysms and pseudoaneurysms can occur in relation to infection. Staphylococcus has been reported to infiltrate the 3 layers of the vessel wall and, in effect, can cause true aneurysm formation, while other organisms tend to more frequently cause pseudoaneurysms [4]. Historically, tuberculosis and syphilis have been common leading causes, however, fungal infections have become the more frequent etiology due to developments in medical therapy [4].

Aspergillus lung infection is most frequently associated with immunocompromised state. Imaging findings can vary from mass-like soft tissue within a lung cavity (aspergilloma) to nodules surrounded by a "halo sign" as seen in angio-invasive aspergillosis. Angio-invasive aspergillosis is characterized by invasion and occlusion of small to medium-sized pulmonary arteries by fungal hyphae [5].

Management of pulmonary artery pseudoaneurysms is variable and tailored based on its etiology, symptomatology, as well as co-existing comorbidities. Historically, symptomatic pulmonary artery pseudoaneurysms have been managed with surgical techniques, such as wedge resection, lobectomy, aneurysmectomy, and pneumonectomy [4]. These interventions are associated with significant risks of complications, especially in patients with pre-existing pulmonary hypertension. Due to increasing availability of endovascular interventions, minimally invasive catheter directed treatments have emerged as a safe and effective technique [4].

In our presented case, the patient failed conservative treatment and continued to have hemoptysis requiring respiratory support. Bronchoscopic interventions were helpful in maintaining airway patency and clarifying the exact site of bleeding, however, not in stabilizing the bleeding. The patient then

presented to interventional radiology and super-selective coil embolization of the pseudoaneurysm was performed. On follow up 1 day later, patient's hemoptysis resolved, and patient was discharged in a stable condition.

Our case demonstrates that minimally invasive endovascular approach to pulmonary artery pseudoaneurysm management can be helpful and, possibly, lifesaving in critically ill patients. Since the pulmonary arteries are end arteries, there is unavoidable devascularization to the lung tissue distal to the embolization. Post embolization angiography (Fig. 5) depicts the very limited peripheral portion of the left lower lobe that lacks contrast opacification highlighting the importance of the selective embolization to prevent from larger areas of pulmonary damage which may have a critical impact in patients with already compromised respiratory function. As opposed to surgical intervention such as lobectomy, this approach also did not preclude patient for repeat interventions if that became necessary due to preservation of the majority of the lung parenchyma.

Conclusion

This is a rare case of a rapidly enlarging pulmonary artery pseudoaneurysm in a very young patient that was successfully managed with a minimally invasive option. While management of asymptomatic and small pulmonary artery pseudoaneurysm can vary, the symptomatic ones require urgent attention and can be successfully managed using minimally

invasive embolization techniques. This is supported by the positive outcome of our patient postembolization who made an uneventful recovery with resolution of hemoptysis in 1 day.

Patient consent

This statement serves as confirmation that the patient provided informed written consent to publish their medical history relevant to this case report.

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