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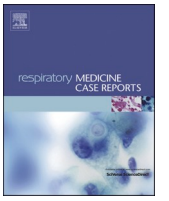

Case Report

Hemoptysis after COVID: Pulmonary artery pseudoaneurysm treated with endovascular coiling

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ABSTRACT

Pulmonary artery pseudoaneurysms are a rare but life-threatening cause of hemoptysis. These are saccular dilatations of the pulmonary artery which do not involve all the three layers of the vessel and are prone to rupture. PAPAs are most commonly associated with infections but may also be caused by vasculitis, lung fibrosis, pulmonary hypertension and a multitude of other causes. CT pulmonary angiography is the diagnostic modality of choice allowing delineation of the PAPA, underlying cause and allowing planning of further management. Although treating the underlying cause is an important part of management, endovascular intervention is required in cases with hemoptysis. Long COVID is an entity that is still under evaluation. It has multisystem involvement. We describe the case of a 30-year-old teacher with hemoptysis after COVID who was diagnosed with a PAPA of segmental branch of left upper lobar pulmonary artery and treated with endovascular embolization with coils and glue. His hemoptysis has stopped since then. He also had pulmonary fibrosis and growth of multidrug resistant *Klebsiella pneumoniae* in sputum. These were treated medically and he has resumed his work as a teacher.

1. Introduction

Long COVID is defined as symptoms of COVID-19 persisting beyond 12 weeks of diagnosis [1]. There is multisystem involvement in long COVID. Pulmonary artery pseudoaneurysms (PAPA) are a rare but life-threatening cause of hemoptysis. Among many other causes, PAPAs have also been associated with infections, lung fibrosis, and vasculitis. All the three complications are seen in long COVID [2]. Here, we describe the case of a 30-year-old teacher with history of COVID-19 presenting with breathlessness and hemoptysis. He had multidrug resistant *Klebsiella pneumoniae* growth in sputum, post COVID pulmonary fibrosis and PAPA. He was treated with coiling for the PAPA and other supportive medical management. At 6-month follow up, he has symptomatic improvement, no hemoptysis, and has resumed work.

2. Case report

A 30-year-old man with history of COVID-19 infection was referred to us for management of hemoptysis. He had no significant medical history prior to being diagnosed with COVID-19 in May 2021 with lung involvement and a CT severity score of 10/25. He had been treated with supplemental oxygen, steroids, anticoagulation, antibiotics, antifungals and antifibrotics and had required bilevel

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non-invasive ventilatory support for respiratory distress. In the first week of July, he developed hemoptysis with about 100 cc blood expectorated per episode and was referred to our tertiary care unit for management. He was tachypneic and tachycardic and had inspiratory crackles on auscultation over all lung fields. On reviewing the CTPA films that the patient had come with (Fig. 1), we noticed a dilated pulmonary artery and cardiomegaly. Interestingly, the diameter of the left pulmonary artery was about 3.2 cm, larger than the right pulmonary artery which measured about 2.2 cm. There was a 2 × 2.1 cm sized non-enhancing thick-walled cavity in the posterior basal segment of the right lower lobe and post COVID fibrosis with multiple areas of traction bronchiectasis, bronchiolectasis, interlobular septal thickening and reticulations. The left pulmonary artery led to a well-defined 2.1 × 1.9 × 1.8 cm isodense nodule in the apico-posterior segment of the left upper lobe with opacification in the pulmonary phase. This was diagnosed to be a pseudoaneurysm arising from a segmental branch of the upper lobar branch of left pulmonary artery (Fig. 2). Echocardiography showed moderate right ventricular dysfunction with an ejection fraction of 60%.

Sputum culture grew multidrug resistant *Klebsiella pneumoniae* and antimicrobial therapy was initiated with colistin. The patient also received hemostatics, supplemental oxygen, antifungals, antifibrotics and supplemental oxygen. The pseudoaneurysm was treated by the interventional radiologists with endovascular coil embolization of the PAPA, and glue embolization of the PAPA and dysplastic artery (Fig. 3). Micro-coils (0.018″) with diameters of 8mm and 10mm, and 14cm length were used. The proximal neck of the feeding artery was short and the aneurysm was arising very close to the origin of the artery. Hence, to avoid the potential risk of proximal coil migration, 50% glue (combination of N-butyl cyanoacrylate and Lipiodol) was used for embolizing the proximal artery. The procedure was performed under local anesthesia. The patient's hemoptysis stopped post coiling. With above medical care, the patient's condition improved. He started maintaining oxygen saturation of 96% at room air and his breathlessness decreased from mMRC grade 4 to mMRC grade 2.

On follow up at six months, the patient had oxygen saturation of 98% at room air, grade 2 mMRC breathlessness and no hemoptysis. A repeat echocardiography showed good biventricular function and a repeat high resolution CT of the lungs showed significant resolution of fibrosis (Fig. 4). The patient had a FVC of 2.13L (60.3% predicted), FEV1 of 1.76 L (62.9% predicted) and FEV1/FVC of 82.41%. On body plethysmography, the patient had a RV of 1.28 L (79.58% predicted), TLC of 3.56L (58.29% predicted) and RV/TLC of 35.97%. This confirmed restrictive pathology. Diffusion study with carbon monoxide had Tlco of 15.88 ml/mmHg/Mi (52% predicted), Va of 2.73 L (45% predicted) and normal KCO 6.54 DLCO/L (130% predicted). The Va/TLC ratio was 76.68. Maximal inspiratory and expiratory pressures generated were 52.78 cmH2O (43% predicted) and 33.78 cm H2O (24% predicted) respectively. The decrease in FVC accompanied by a decrease in TLC is suggestive of a restrictive disorder. The marginal increase in RV/TLC can be explained by a decrease in the FRC and TLC. A decrease in the MIP and MEP may suggest some residual muscle weakness. He continues to be enrolled in the pulmonary rehabilitation programme and has resumed his work of teaching school students.

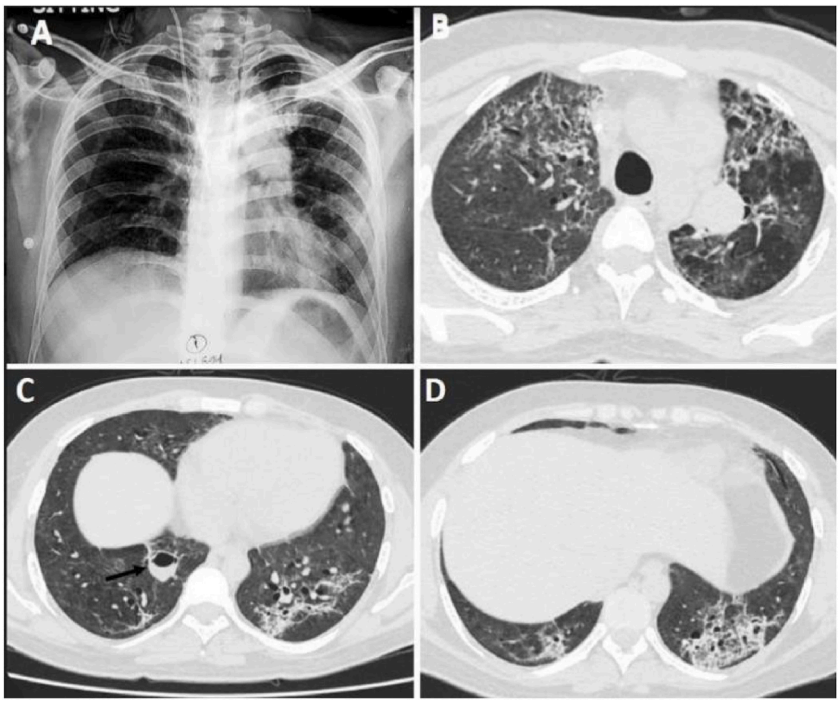

Fig. 1. Frontal chest radiograph (A) shows reticular opacities in both upper zones. Axial sections of HRCT Chest (B, C, D) show reticular opacities in both lung fields with septal thickening and multiple areas of traction bronchiectasis. A thin-walled cavity is seen the right lower lobe (black arrow in C).

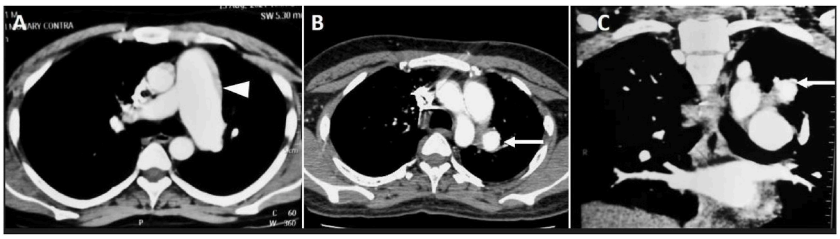

Fig. 2. Axial section of CT Pulmonary Angiogram (A) shows dilated main and left pulmonary artery. Axial (B) and coronal (C) sections of CTPA shows pulmonary artery pseudoaneurysm arising in the left upper lobe.

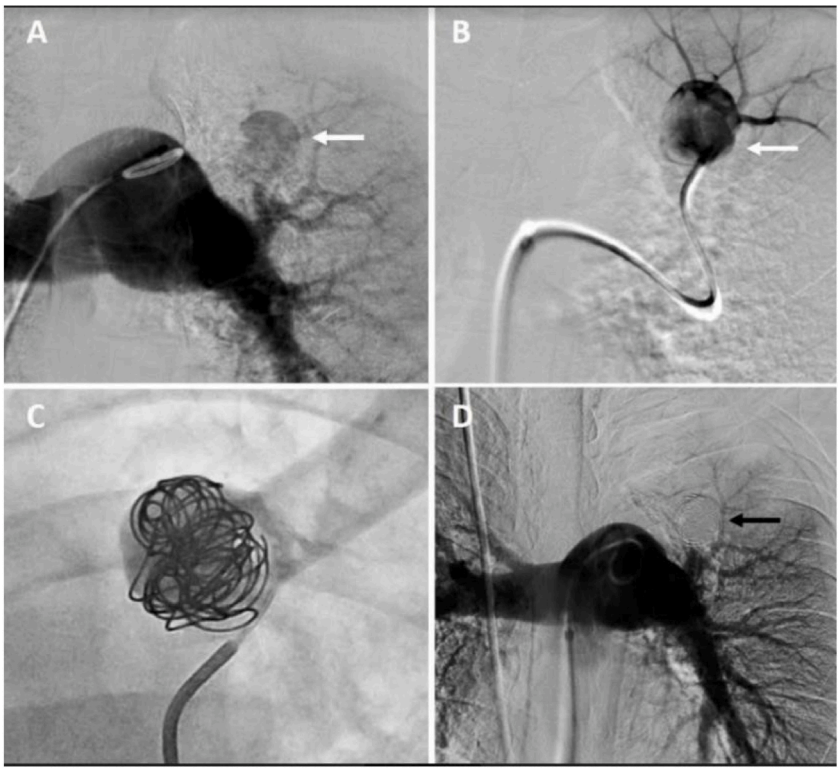

Fig. 3. (A) Catheter Pulmonary Angiogram shows PAPA (white arrows) arising from a segmental branch of the left upper lobe. (B) Selective angiogram performed after cannulation of the abnormal feeding artery of the PAPA. (C) Endovascular coil embolization of the PAPA, with glue embolization of the PAPA and dysplastic artery. (D) Check pulmonary angiogram shows complete occlusion of the aneurysm.

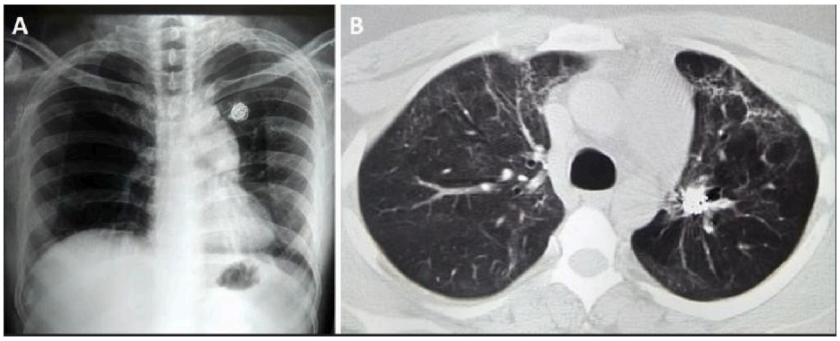

Fig. 4. Follow up chest radiograph (A) and axial HRCT Chest (B) shows metallic coils in left upper lobe with reduced lung fibrosis.

3. Discussion

While a true pulmonary artery aneurysm (PAA) is described as focal dilatation of all three layers of the pulmonary artery, PAPAs do not involve all three layers and are prone to rupture [3]. PAPAs therefore have a high mortality rate, reaching 50% in untreated cases [2]. Proximal PAPAs are those arising from the pulmonary trunk and may be associated with pulmonary hypertension. PAPAs situated on an intrapulmonary artery are classified as peripheral and are life-threatening. Peripheral PAPAs are more common [4]. PAPAs are commonly solitary with multiple PAPAs usually associated with endocarditis and metastatic disease to the lung. PAPAs are further classified angiographically into types A-D. Type A PAPAs are visualized at nonselective pulmonary angiography and therefore are assumed to have a patent feeding pulmonary artery without significant bronchopulmonary shunting. Type B PAPAs are those visualized at selective segmental or subsegmental pulmonary angiography. Type C PAPAs are apparent at bronchial and non-bronchial systemic collateral arterial angiography due to bronchopulmonary arterial shunt but not at selective pulmonary angiography. PAPAs apparent at pulmonary CT angiography but not at catheter-directed angiography are classified as type D [5].

PAPAs can be congenital or acquired. Infections are the commonest cause of PAPAs.

Tuberculosis and syphilis were the commonest infections thus far but with effective chemotherapy for both, other pyogenic organisms like *Staphylococcus aureus*, Klebsiella and Streptococcus pyogenes and fungi like Mucor, Aspergillus and Candida are becoming frequent. It is hypothesized that repeated seeding of the endovascular wall in an infection weakens the wall from inside, finally leading to an aneurysmal dilatation. Other causes of PAPAs have been described in several case reports. These include trauma, iatrogenic due to catheters, lung cancers, pulmonary embolism, vasculitis, traction bronchiectasis and pulmonary fibrosis among others [2,6].

COVID-19 has been linked to a vasculitis-like process due to upregulation of proinflammatory mediators and endothelial damage and dysfunction. Considering that vasculitides like Behcet's syndrome have been associated with PAPAs, the mechanism underlying PAPAs in COVID-19 could be vasculitis and needs further investigation. Mucormycosis is another cause of PAPAs in COVID-19 survivors [7–9].

Patients with PAPA commonly present with hemoptysis and may also have chest pain. They may be hypoxic but this may be due to the underlying condition. Chest radiographs may show round, well-circumscribed nodules or hilar enlargements or even focal consolidation. CT angiography is the current modality of choice for detection of PAPAs. CT angiography may reveal enlarged pulmonary arteries with mural thrombus in a proximal PAP. Peripheral PAPAs usually present as saccular distal aneurysms. Associated pulmonary hypertension can be diagnosed with enlarged right ventricle and pulmonary trunk with mosaic perfusion. CT angiography can also identify the underlying cause of PAPA [4].

Medical therapy to treat the underlying cause of the PAPA is an important step in treatment.

However, definitive management with endovascular intervention or surgery is necessary. Surgical options for treating PAPAs include aneurysmorrhaphy, lobectomy, aneurysmectomy and pneumonectomy. However, surgical resection is fraught with risks. These patients are often very ill, have sepsis, or may have pulmonary hypertension increasing the risk of surgery. Therefore, when the PAPA is amenable to endovascular treatment, it is the first line therapy of choice. Central fusiform aneurysms require surgical management [10,11].

Coil embolization is the preferred management option. This is because the intra-saccular coils allow preservation of pulmonary arteries distal to the PAPA. Embolization is chosen as the therapeutic option when there is an increased risk of rupture of the PAPA. However, in cases of proximal embolization, the aneurysmal sac may continue to be perfused by distal bronchopulmonary anastomoses and therefore requires ruling out a shunt. Vascular plugs have also been used as an alternative to coils. There are reports of successful glue embolization with n-butyl cyanoacrylate (NBCA) [10,12].

Post COVID pulmonary fibrosis is an entity that is under study. Early reports suggest that this may be reversible. Similarly, there are reports of diaphragmatic palsy in post-acute sequelae of COVID-19 and muscle weakness in long COVID [13–16].

In conclusion, clinicians must have a high index of suspicion for vascular pathologies in cases of hemoptysis. Pulmonary artery dilatation is not always only pulmonary hypertension and widely differing diameters of the left and right pulmonary arteries may provide a clue to presence of unilateral pathology and aneurysms. Several factors may contribute to the development of PAPAs in patients with history of COVID-19. Timely endovascular intervention is the cornerstone of managing PAPAs.

Declaration of interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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