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CASE REPORT

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Pulmonary pseudoaneurysm in the setting of concurrent COVID-19 and pulmonary mucormycosis: A rare case report

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Key Clinical Message

This case illustrates the possible danger of PAP emergence in individuals with a confluence of conditions capable of inducing vascular impairment, like COVID-19, pulmonary mucormycosis (PM), and diabetes.

Abstract

Pulmonary mucormycosis (PM) is a highly lethal invasive infection. It is a rare complication of COVID-19 and is associated with a high mortality rate. Pulmonary pseudoaneurysm (PAP) is a severe manifestation of this condition, often resulting in death. Management involves endovascular therapy followed by surgery and appropriate antifungal treatment.

KEYWORDS

case report, COVID-19, hemoptysis, pulmonary mucormycosis, pulmonary pseudoaneurysm

1 | INTRODUCTION

Pulmonary mucormycosis (PM) is recognized as an invasive infection characterized by a high mortality rate, posing challenges in its management and treatment. PM accounts for approximately 10%–20% of all mucormycosis cases, with less than 10% occurring in individuals with COVID-19.[1,2] Pulmonary pseudoaneurysm (PAP), an uncommon occurrence resulting from the 2019 coronavirus disease (COVID-19) and PM, was first documented by Dantis et al. in 2021.[1] This manifestation is particularly severe and often fatal when accompanied by hemoptysis. The recommended approach to managing this complication involves endovascular therapy as a temporary measure before definitive surgery.[2] Additionally, the administration of appropriate antifungal treatment is crucial in determining the patient's outcome.[3] In this report, we present an extremely uncommon occurrence of pulmonary pseudoaneurysm alongside coexisting PM and COVID-19 infections.

2 | CASE PRESENTATION

A 41-year-old male patient with a history of COVID-19 presented to our hospital after experiencing episodes of hemoptysis over a 10-day period. The blood loss experienced was significant and clotted. The patient did not exhibit symptoms such as fever, dyspnea, chest pain, palpitations, or syncopal episodes. Additionally, he had a medical history of type 2 diabetes, which required insulin treatment. In the past, the patient had undergone lithotripsy and had undergone surgery to repair his right knee meniscus. The occurrence of hemoptysis came

approximately 1 month after the diagnosis of COVID-19 was confirmed using the SARS-CoV-RNA transcription-mediated amplification test and a chest computed tomography (CT) scan. The CT scan revealed widespread infiltrations in the lungs that were consistent with COVID-19, but no aneurysms were observed at that time (Figure 1). The patient had been under home isolation and had been prescribed systemic glucocorticoids (specifically methylprednisolone) for 1 week as part of the management plan.

One month following the onset of COVID-19, the patient experienced a dry cough, followed by episodes of hemoptysis, with approximately 100 mL of blood being expelled per episode. As a result, the patient was referred to our department for the management of symptoms. During the chest examination, it was observed that the chest was symmetrical with no external deformities. Subsequent assessment revealed normal breath sounds upon auscultation, and no palpable masses were detected. A complete blood count (CBC) indicated a hemoglobin level of 6.4 g/dL, while liver and renal functions were within normal range. To further investigate the condition, a computed tomographic pulmonary angiography (CTPA) was conducted. The scan revealed the presence of a pseudoaneurysm, measuring 3 cm in length and 3.8 cm in diameter, in the pulmonary artery of the right lower lobe. This pseudoaneurysm originated from a branch of the right descending pulmonary artery and was accompanied by basal segmental consolidation and adjacent minimal pleural effusion. Venous dilatation was also observed in the right lower lobe, with slight right lateral blunting, although there were no clear signs of pulmonary embolism (Figure 2). Based on these findings, a preliminary diagnosis of pulmonary artery pseudoaneurysm was suggested. Bacterial growth was not observed in the patient's sputum, and acid-fast bacilli tests yielded negative results in three consecutive instances. After rectifying the decreased hemoglobin level through a blood transfusion and achieving a hemoglobin level of 9 g/dL, a bronchoscopy procedure was carried out.

It has been determined that a rigid bronchoscopy will be conducted for the purpose of evaluation and diagnosis. General anesthesia was administered, and thrombi were discovered protruding from the intermediate bronchus toward the right main bronchus during the procedure. Due to the potential risk of sudden bleeding, the orifices of the upper and intermediate lobes were deliberately left throughout the operation.

A decision was made to proceed with a right thoracotomy. Adhesions were released, and the right lower lobe was found to be filled with thrombi and blood, rendering it non-functional for respiration. The lower pulmonary artery and vein were isolated and securely tied off. Subsequently, the lower lobe bronchus was meticulously dissected and sutured separately. Bronchoscopy was performed concurrently with the surgical procedure, confirming that the upper and middle lobe bronchi were open and functioning adequately. To facilitate drainage, a chest tube was inserted, and the surgical intervention proceeded smoothly, devoid of any complications or unfavorable outcomes both during and after the operation.

Upon gross examination, the resected right inferior lobe was found to weigh 450 g and measure 20 × 17 × 15 cm. Subsequent sectioning of the tissue revealed condensed, edematous, and congested pulmonary tissue. There were areas of dark, friable consistency with hemorrhage and thrombi, as well as a small segment exhibiting spongiosis (Figures 3 and 4). The pathological examination indicated widespread inflammatory changes in the pulmonary tissue, with alveolar spaces filled with proteinaceous material and areas displaying hemorrhagic ischemic changes. Additionally, thick fungal hyphae with angioinvasion were observed suggesting infection by mucormycosis (Figure 5). Reactive changes were observed in the lymph nodes. The diagnosis confirmed a COVID-19 viral infection complicated by infarction and focal infection of mucormycosis. After that the patient was put on liposomal amphotericin-B (beginning with 3 mg/kg until discharge).

During the post-operative evaluation, it was noted that the patient's hemoptysis had ceased. The post-operative

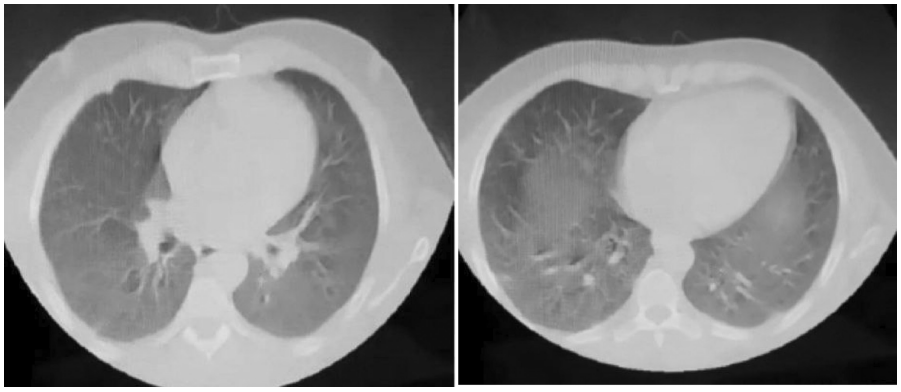

FIGURE 1 CT scan revealing widespread infiltrations in the lungs that were consistent with COVID-19.

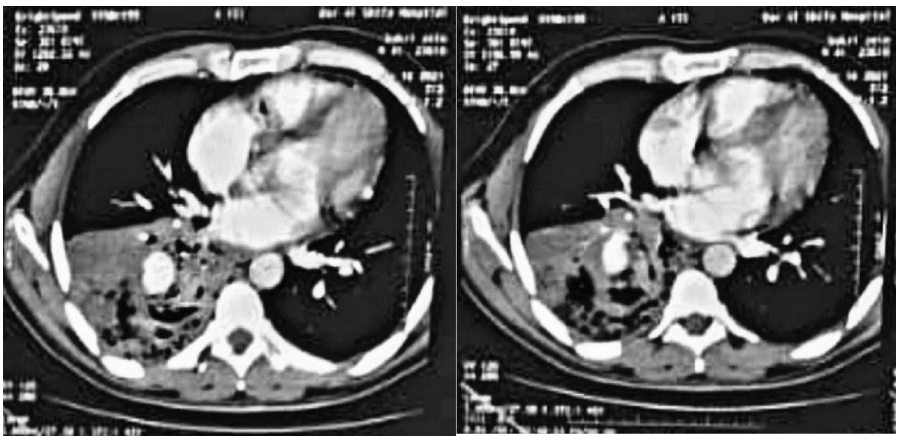

FIGURE 2 A CTPA scan revealed a 3 cm long and 3.8 cm wide pseudoaneurysm in the right lower lobe pulmonary artery. It was accompanied by consolidation, pleural effusion, and venous dilatation in the same area, with no clear signs of pulmonary embolism.

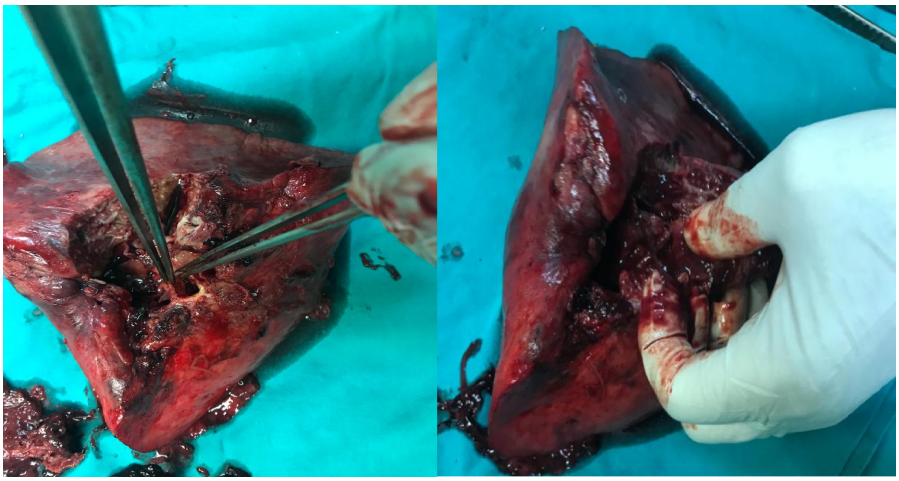

FIGURE 3 Gross photograph showing the resected right inferior lobe showing the lumen of the pseudoaneurysm.

recovery process was meticulously managed, with emphasis placed on effective pain management, early mobilization, respiratory care, and adequate nutritional support. Continuous monitoring of the patient was conducted throughout the recovery period to facilitate proper healing and minimize the potential for complications. The patient responded favorably to the post-operative care, and the recovery process transpired without any significant incidents. Following the removal of the chest tube, the patient was discharged in a stable condition with a normal chest X-ray, 6 days after the operation (Figure 6). A three-month follow-up period ensued, during which no recurrence of hemoptysis or post-operative complications were observed. The patient's overall health was deemed satisfactory, as there were no notable reports of pain or discomfort.

3 | DISCUSSION

Pulmonary artery pseudoaneurysm is a condition characterized by the dilation of a branch of the pulmonary artery, primarily affecting the outer layer of the arterial wall.[4] This uncommon occurrence is typically observed in patients who have undergone bronchial or pulmonary angiography, leading to the development of hemoptysis. The estimated prevalence of this condition in such patients is approximately 11%.[5] Various risk factors contribute to its occurrence, including traumatic injury, malignancies, and chronic inflammation. Infection serves as the primary cause of pulmonary artery pseudoaneurysms, accounting for over 33% of cases.[4] The formation of pseudoaneurysms arises from vessel wall inflammation due to pneumonia, septic emboli, invasive organisms, or tumors, resulting in weakened arterial walls and the subsequent formation of pseudoaneurysms.[6]

Recent research has revealed the association of COVID-19 with diffuse alveolar damage, immunothrombi in the pulmonary vasculature, and inflammatory and vasculitic effects on the skin, pulmonary vasculature, and a Kawasaki-like disease phenomenon.[7] Additionally, a concerning increase in the incidence of PM has been observed globally, particularly among individuals who have previously contracted COVID-19.[3,8] The occurrence rate of PM varies from 0.005 to 1.7 per million population.[9] Although PM is rare, it should not be underestimated, particularly in immunosuppressed patients, as it can also serve as a causative factor for the development of PAP.[10] PM is known to invade blood vessels

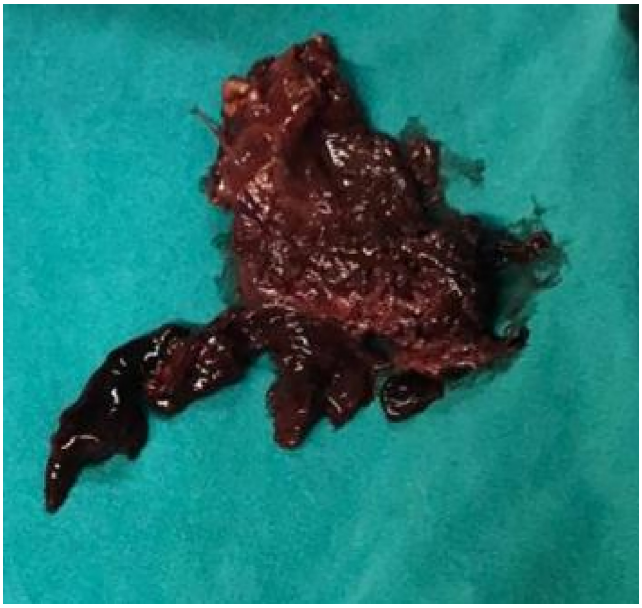

FIGURE 4 The gross photograph presented exhibits the thrombi that have been excised from the lower lobe of the right lung.

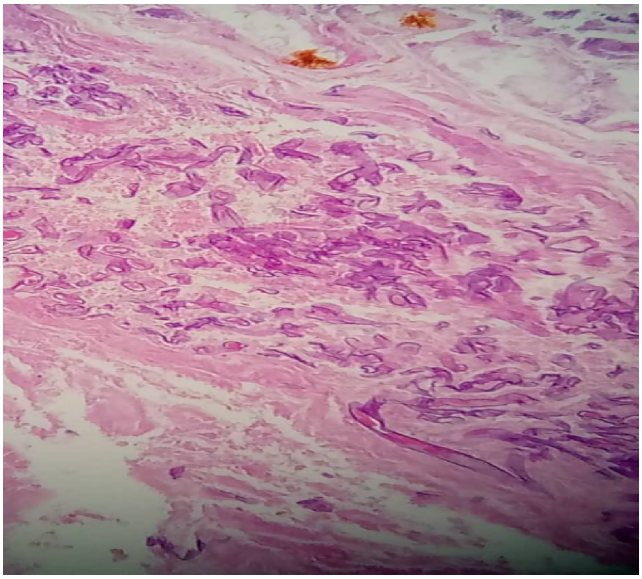

FIGURE 5 Microscopy showing thick fungal hyphae suggestive of mucormycosis.

through its hyphae, leading to complications such as hemorrhage, thrombosis, infarction, and tissue necrosis.[11] Individuals with compromised immune systems are at an elevated susceptibility to acquiring mucormycosis infection, particularly those who have recovered from COVID-19, individuals undergoing immunosuppressive therapy, individuals on long-term corticosteroids, or diabetes with uncontrolled hyperglycemia.[12] Glucocorticoids inhibit the functioning of leukocytes during inflammation and suppress the humoral

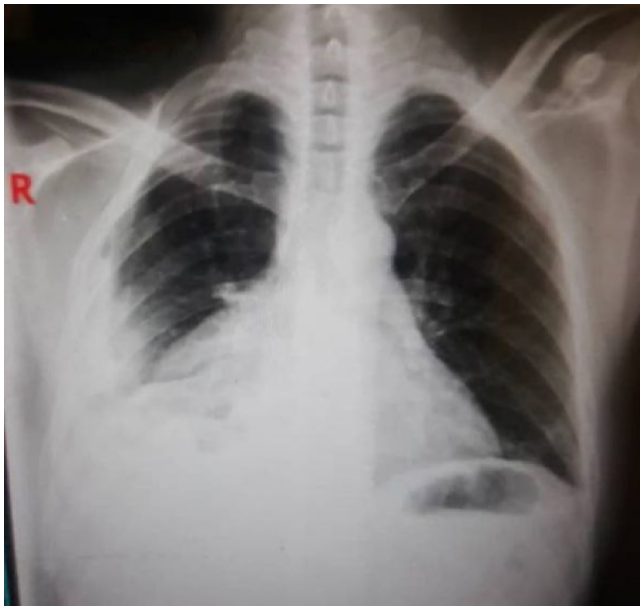

FIGURE 6 A CXR performed 4 days post-surgery showed normal findings.

factors associated with it.[13] In our specific case, the patient had previously contracted COVID-19 and was treated with glucocorticoids, which can induce immunosuppression leading to PM. The patient also had type 2 diabetes, which can further impair immune function as stated in the literature especially in the presence of hyperglycemia which means that the patient might had a chronic PM previous to COVID-19 infection.[14,15] Considering these factors, such as immunosuppression and the inflammatory condition of the vasculature, we hypothesize that the combination of coronavirus disease, pulmonary mucormycosis, and diabetes poses a substantial risk for the development of PAPs, A case series involving five patients who had both coronavirus and pulmonary mucormycosis revealed that all of them had diabetes, and three were treated with glucocorticoids. Among all five patients, PAPs emerged, which supports our hypothesis.[2] However, in our case, it is difficult to determine whether the presence of PM was a result of COVID-19 infection and glucocorticoid use, or if it was a chronic condition related to diabetes.

Based on a study conducted on 13 patients, it has been observed that PAP manifests with symptoms such as dyspnea, chest pain, and hypoxia. However, the most prevalent indication is the presence of hemoptysis, which can vary in severity from being life-threatening and potentially fatal to being incidentally detected in radiographic examinations. Therefore, PAPs can pose a significant risk to individuals or progress silently as an enlarging lesion.[16] To be categorized as massive hemoptysis, pulmonary hemorrhage requires the expectoration a significant amount of blood within a day. If left untreated, mortality rates can rise to approximately 50%.[17] In our patient, the hemoglobin level dropped

significantly to 6.4g/dL, indicating a severe hemorrhage. While hemoptysis is an uncommon symptom in coronavirus disease,[18] its presence in a COVID-19 patient should lead us to consider the possibility of an associated infection with vascular dysfunction properties.

Computed tomography pulmonary angiography (CTPA) is the preferred diagnostic test for detecting localized dilation of a pulmonary artery branch.[5,19] Therefore, a CTPA was performed in this particular case. Given their atypical presentation, pulmonary artery pseudoaneurysms (PAPs) often go unnoticed by radiologists, with up to 46% of cases being initially missed on CT scans.[5] This delayed diagnosis can result in severe complications such as PAP rupture and massive hemoptysis, which carries a 50% mortality rate if left untreated.[20] Hence, it is crucial to maintain a high level of suspicion and carefully consider the patient's clinical history to ensure a favorable outcome. Additionally, CTPA aids in identifying suitable candidates for potential endovascular interventions.[10] Microorganism demonstration in tissue is the gold standard for confirming a clinical diagnosis of PM. However, this can be challenging and is often not feasible before surgery.[10] Focal consolidations observed on imaging can be differentially diagnosed as pulmonary thromboembolism, superimposed pneumonia, post-COVID fibrosis, or arterial pseudoaneurysm.[21,22]

The preferred method for managing PAPs is endovascular treatment, which includes coil embolization or stent placement.[5,19,23] This approach offers the advantage of preserving blood flow beyond the PAP while carrying a lower risk of complications compared to surgery.[21] However, in cases where there is a rupture of the PAP sac, such as the one described here, early surgical intervention is necessary to ensure patient survival.[10] In addition to that, endovascular equipments are not available in our hospital due to lack of the necessary resources and financial means. Surgical interventions may involve procedures like pneumonectomy, aneurysmectomy, pulmonary artery arteriorrhaphy, and lobectomy.[10] Subsequent pathological investigations confirmed the presence of a localized mucormycosis infection. Successful management of this condition requires the administration of antifungal drugs, removal of all infected tissues, and the implementation of other supplementary therapies.[24] Therefore, amphotericin was initiated, and complete removal of the infected tissue was already performed during the aforementioned surgical intervention.

4 | CONCLUSION

In conclusion, the presented case highlights the potential risk of developing PAPs in patients with a combination of diseases that has potential to cause vascular dysfunction such as COVID-19, pulmonary mucormycosis (PM), and diabetes. Early diagnosis of PAPs is crucial to prevent severe complications such as rupture and massive hemoptysis, which can lead to high mortality rates if untreated. CTPA is the preferred diagnostic test for PAPs, although initial CT scans may miss cases, emphasizing the need for suspicion and careful consideration of the patient's clinical history. Endovascular treatment, such as coil embolization or stent placement, is the preferred management approach, preserving blood flow and minimizing complications. However, in cases of PAP rupture, early surgical intervention becomes necessary for patient survival. Successful management of PM involves antifungal drug administration, removal of infected tissues, and supportive therapies.

AUTHOR CONTRIBUTIONS

Hussein Hamdar: Data curation; writing – original draft; writing – review and editing. **Ali Alakbar Nahle:** Writing – original draft; writing – review and editing. **Rida Jaber:** Writing – original draft. **Hadi Salame:** Writing – original draft. **Amjad Sikaria:** Resources; writing – review and editing. **Younes Souleiman:** Supervision; visualization.

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None.

CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

Data are available upon request due to privacy/ethical restrictions. The data that support the findings of this study are available upon request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

CONSENT STATEMENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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