

Case Report

Spontaneous alveolo-pleural fistula in a case of COVID-19 pneumonia- challenges and solutions: case report

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ABSTRACT

COVID-19 pneumonia has demonstrated a wide spectrum of clinical presentations that have yet to be completely uncovered. As this pandemic progresses, uncommon presentations of this disease have come into light. Development of broncho/alveolo-pleural fistula in a patient with COVID-19 pneumonia is a rare phenomenon with only 4 cases reported in literature till date. A 61-year-old gentleman presented to the emergency department with fever, cough, and shortness of breath. His initial chest X-ray was suggestive of a viral pneumonia that was later confirmed to be due to COVID-19. The patient was put on non-invasive ventilator support and treated with empirical antibiotics, glucocorticoids, anti-viral medications and convalescent plasma therapy. Four weeks into the patient's hospital course, his vital parameters suddenly deteriorated with a subsequent chest X-ray showing a tension pneumothorax, for which a chest tube insertion was done. However, when the air leak did not resolve by the 3rd day, a chest computed tomography (CT) was planned which showed a spontaneous alveolo-pleural fistula (APF). The patient was managed with conservative treatment using negative suction applied to an underwater seal, had his chest tube removed 10 days later and was discharged subsequently. Spontaneous fistulisation between broncho-alveolar tree and pleura can occur rarely in patients with COVID-19 pneumonia and can be managed using underwater seal with negative suction, insertion of endobronchial valves or surgical closure, and needs to be individualised.

Keywords: COVID-19, Necrotizing pneumonia, Persistent air-leak, Alveolo-pleural fistula

INTRODUCTION

COVID-19 infection can present with a myriad of clinical signs and symptoms, ranging from mild asymptomatic disease to fulminant pulmonary involvement and as this pandemic progresses, uncommon presentations of this disease have come into light.

Recent evidence has suggested the occurrence of spontaneous pneumothorax in the context of COVID-19 pneumonia.¹ Here, we report a case of spontaneous pneumothorax with a persistent air-leak, which was found to be due to an alveolo-pleural fistula, a rare complication seen with COVID-19 pneumonia. To the best of our

knowledge, only 4 cases of broncho/alveolo-pleural fistula in a patient with COVID-19 pneumonia have been reported in literature till date.

CASE REPORT

A 61-year-old gentleman with no significant past medical history (non-smoker with no history of any pre-existing lung disease or pulmonary Kochs) presented to the emergency department with fever, dry cough and shortness of breath for 1 week. On examination, he was found to be afebrile, tachycardic with a heart rate of 112 bpm, tachypnoeic with a respiratory rate of 28 breaths/min and hypoxic with a saturation of peripheral

oxygen (SpO₂) of 77% on room air. Respiratory examination revealed crepitations in bilateral lung fields. Chest X-ray revealed bilateral pneumonitis suggestive of viral pneumonia (Figure 1).

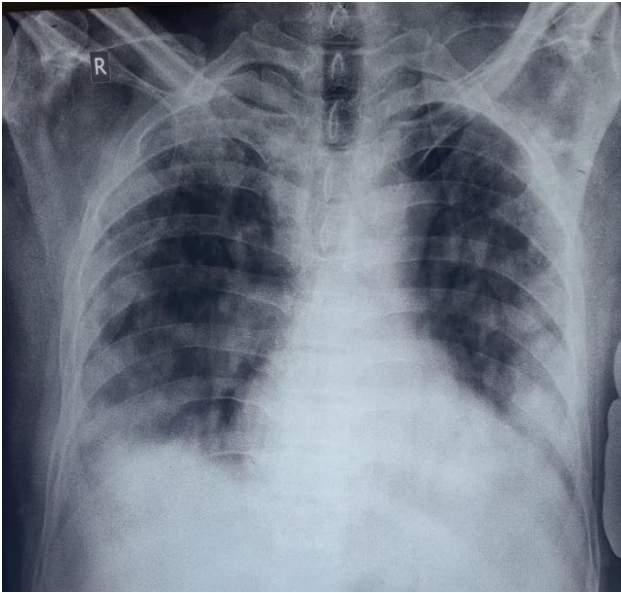


Figure 1: Initial chest X-ray of bilateral pneumonitis.

Laboratory analysis noted an elevated white blood cell count of 16,300/mm³ (absolute lymphocyte count= 815/mm³), elevated C-reactive protein (CRP) and D-dimer levels (12.6 mg/dL and 1970 ng/mL respectively). A real-time reverse transcriptase polymerase chain reaction (RT-PCR) test for SARS-CoV-2 RNA was performed which came out to be positive.

Patient was admitted in an intensive care unit and put on non-invasive ventilator support and was managed as per ICU protocols of our institute. Initial treatment included empirical antibiotics, low molecular weight heparin and a steroid course with dexamethasone. Convalescent plasma therapy and a course of anti-viral drug, remdesivir was also given to the patient.

The patient's clinical picture started improving and by the end of third week, his ventilator support had been de-escalated and the patient was shifted to oxygen supplementation by a non-rebreathing face mask.

However, a week later, the patient developed complaints of sudden onset right-sided chest pain with breathlessness. On examination, he was found to be tachypneic with reduced air entry on the right side of the chest. An urgent bedside chest X-ray revealed a large right-sided pneumothorax with collapse of the underlying lung, with mediastinal shift towards the left side. A 30-French size chest tube (ICD) was inserted bedside by the surgical team. Post-ICD insertion chest film showed a persistent right pneumothorax with surgical emphysema at the site of chest tube insertion (Figure 2).

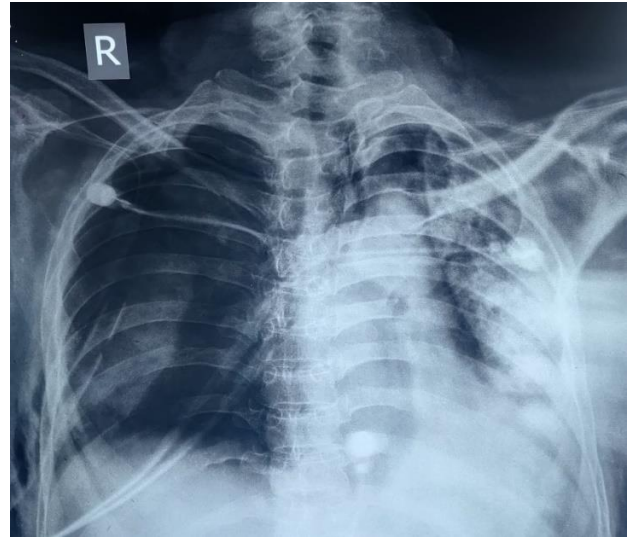


Figure 2: Chest X-ray of persistent right-sided pneumothorax with mediastinal shift towards the left side. Note the surgical emphysema at the site of chest tube insertion.

When the air-leak did not resolve by the third day, a CT chest was planned for further evaluation. It noted a massive right-sided pneumothorax with extensive subcutaneous emphysema along with a suspicious breach in the visceral pleura of the right lung, raising the possibility of a spontaneous alveolo-pleural fistula (Figure 3).

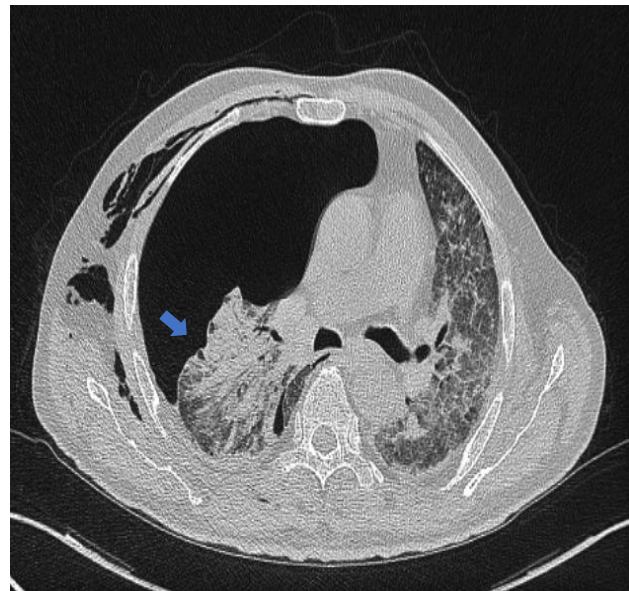


Figure 3: Axial section from CT chest; a massive right-sided pneumothorax with extensive subcutaneous emphysema. A suspicious breach is seen in the visceral pleura of the right lung (blue arrow) raising the possibility of an alveolo-pleural fistula.

The cardiothoracic department and pulmonology were consulted and conservative management was advised by

using negative suction applied to the underwater seal. Negative suction equivalent to -10 cm of water was applied to the ICD. Over the next few days, the air-leak reduced gradually and column movement in the ICD ceased. Subsequent chest X-ray showed an expanded right lung with no residual pneumothorax. The chest tube was removed 10 days after the application of negative suction and the patient was finally discharged in a stable condition after about 60 days of hospitalization.

DISCUSSION

Broncho/alveolo-pleural fistula refers to the presence of an abnormal connection between the broncho-alveolar tree and the pleural space. It causes increased morbidity and mortality to the patient. Though most frequently seen as a post-operative complication of pulmonary resection, several case reports of fistula caused secondary to a necrotizing pneumonia have been published in literature with commonly implicated organisms being *Pneumococcus*, *Staphylococcus aureus* and *Acinetobacter baumannii*. Interestingly, some similarities seem to be present in the histopathological picture seen in these necrotizing pneumonias and COVID-19 pneumonia.

In a retrospective study of 112 children, Hsieh et al concluded that most of the children with necrotizing pneumococcal pneumonia complicated with broncho-pleural fistula, had the same pathologic features as those seen in pulmonary gangrene, which develops due to ischemia and pulmonary infarction secondary to arteritis and vascular thrombosis in the pulmonary vessels supplying that part of lung.² Similar evidence of thrombotic micro-angiopathy (including capillary, arteriolar or venous thrombus formation) and hemorrhagic pulmonary infarction was found in a post-mortem lung-biopsy study conducted in patients infected with SARS-CoV-2 by Beigee et al and Lax et al.^{3,4}

We found only a few case reports describing this communication between the broncho-alveolar tree and pleura, and only a single case report describing an alveolo-pleural fistula in the setting of COVID-19 infection.⁵⁻⁸ The first line of management of such a fistula is ICD insertion with an underwater seal, failing which negative suction application might be required, as was seen in our case. Alveolo-pleural fistula should usually respond without requirement of any further intervention. However, if the air leak fails to resolve this way, placement of an endobronchial valve (EBV) can help in reducing the air leak, which may then gradually resolve with the water seal.⁶ This is especially beneficial in patients with COVID-19-related acute respiratory distress syndrome (ARDS) who require positive pressure ventilation, as it reduces volume loss through the fistula, and facilitates ventilator weaning.⁵

Surgical management is reserved for patients with a broncho-pleural fistula (BPF) or a large alveolo-pleural fistula and where surgical closure of the same has high

likelihood of failure. Placik et al reported the occurrence of a large BPF secondary to mucormycosis in a 49-year-old gentleman suffering from COVID-19 pneumonia.⁷ Surgical debridement and repair of the BPF was done. In a similar case reported by Peeters et al, where emergency right thoracotomy had to be performed in a 36-year-old woman, a central BPF was discovered intra-operatively. It was treated with extensive decortication and salvage lobectomy, as closure was not feasible owing to the size of the fistula.⁸

CONCLUSION

Physicians treating COVID-19 patients should be aware of the entity of spontaneous fistulisation between broncho-alveolar tree and pleura and their management needs to be individualised.

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