**Introduction**

Fistula is an abnormal anatomical communication between an organ, vessel, or intestine and another structure. Bronchopleural fistula (BPF) is a common entity encountered by pulmonologists where there is an abnormal communication between a bronchus and the pleural space due to various reasons like rupture of lung abscess, necrotising pneumonia, or tuberculosis. Prevalence of BPF in cases of COVID-19 is unknown, so is the aetiology. Excessive use of mechanical ventilation, longer duration of steroid use, and secondary bacterial infections could be the possible explanations; however, the role of pre-existing aetiologies like tuberculosis or chronic lung diseases cannot be ruled out. COVID is an ongoing pandemic which is initially managed by primary care physicians. They, thus should have a gross knowledge about these emerging complications, so that they can diagnose them easily and try to manage or refer at the appropriate time. They should act rationally in prescribing steroids and titrating the parameters of positive pressure ventilation. We hereby present a series of six cases of COVID lung with BPF that had different aetiologies with various patterns of presentations and outcomes.

**Keywords:** Bronchial fistula, COVID-19, noninvasive ventilation, pneumothorax

**Case 1**

A 31-year-old male patient was admitted to the intensive care unit (ICU) with fever and breathlessness. He had no comorbidities or addictions. The patient had a computed tomography (CT) severity score of 20. He was managed with non-invasive ventilation (NIV) support. The routine investigations showed a high total leucocyte count (14500/mm$^3$), CRP (25.4 mg/ml), and d-dimer (1.1 mcg/ml). He had sudden increase in the oxygen requirement (FiO₂). The chest radiograph revealed a right sided pneumothorax for which inter-costal drainage (ICTD) was inserted. Persistent pneumothorax with bronchopleural fistula in COVID lung – An emerging challenge

Suman K. Jagaty¹, Debasis Behera¹, Saurabh Gupta², Saswat Subhankar¹, Chinnari M. Rao¹, Maram R. Jeeshitha¹

Departments of ¹Respiratory Medicine and ²Critical Care Medicine, Kalinga Institute of Medical Sciences, Bhubaneswar, Odisha, India

**Abstract**

Bronchopleural fistula (BPF) is an uncommon entity in patients with coronavirus disease 2019 (COVID-19). It may arise due to various causes ranging from the disease itself to complications of treatment. If left untreated, it may increase the morbidity, hospital stay, and adversely affect the outcome. We hereby present a series of cases with a persistent pneumothorax and associated BPF due to varying aetiologies. While three of our cases developed a pneumothorax while on non-invasive ventilation, other three were on oxygen therapy. One patient developed a spontaneous pyopneumothorax and septicemia and succumbed to the complications. Another patient on non-invasive ventilation died due to complications of pregnancy. The management of each case varied depending on their clinical presentation.

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done [Image 1(a)]. The patient, however, had a persistent air leak although he showed clinical improvement. Later, he was discharged with home oxygen support. He was re-admitted after five days with complaints of fever and breathlessness. On presentation, he had a pulse rate of 112/min and oxygen saturation (SpO2) of 97% with oxygen of 4 litres/min.; his temperature was 100.1°F. His CRP was 96 IU/L at presentation which gradually increased to 314 over 4 days. All other routine blood investigations were within the normal limits. CT of thorax showed right moderate hydropneumothorax with diffuse ground glass opacities in both lungs [Image 1(b)]. There was no spontaneous closure of the fistula; pleural fluid culture grew *Klebsiella* sps. which was sensitive to tigecycline. Patient improved clinically with antibiotics, but BPF persisted for which patient was transferred to higher centre for surgical management. The patient was doing fine at follow-up.

**Case 2**

A 38-year-old male patient with no known comorbidities, non-smoker and with no previous medical or surgical history was admitted to the ICU with complaints of breathlessness for four days. He was put on NIV support. All his routine parameters were within normal limits. He developed a right-sided pneumothorax during hospital stay [Image 2(a)]. Intercostal chest tube was inserted and then it was found to have BPF. The patient had to be discharged on request with oxygen support. Five days after discharge, he presented to the emergency room with complaints of fever and shortness of breath. On presentation, he had a pulse rate of 122 bpm, respiratory rate of 28 cycles/min and maintained a SpO2 of 100% with 5 litres/min of oxygen. His laboratory parameters were insignificant except for elevated CRP (386 IU/L). Air-leak was persistent. CT thorax showed right moderate pneumothorax with patchy consolidation and multiple intraparenchymal cystic changes in left lung and right lower lobe [Image 2(b)]. Autologous blood instillation was done in view of persistent pneumothorax with BPF as patient refused any surgical interventions. The patient gradually improved, and air-leak subsided completely. Repeat chest radiograph showed complete lung expansion within 10 days and intercostal tube was removed [Image 2(c)]. The patient was discharged with stable hemodynamic status.

**Case 3**

A 36-year-old male patient, non-smoker with no known comorbidities was referred from a COVID hospital due to worsening dyspnoea and increase in oxygen requirement. On presentation patient was conscious and oriented with a pulse rate of 130 bpm, respiratory rate of 32 cycles/min, and SpO2 of 88% with 15 litres/min of oxygen. Peripheral cyanosis was present. His laboratory parameters showed elevated inflammatory markers (CRP-138 mg/dl, serum ferritin-855 ng/ml, d-dimer-1.05 mcg/ml) and total leukocyte count (22300 cells/mm³). Chest X-ray showed a right sided pneumothorax [Image 3(a)]; CT thorax was suggestive of a cavitary lesion in superior segment of right lower lobe and right-sided hydro-pneumothorax [Image 3(b)]. Intercostal tube (ICT) was inserted and post-procedure BPF was confirmed by the presence of a persistent air-leak. The patient was treated with injectable antibiotics, steroids, and anticoagulants. The pleural fluid was positive for *Mycobacterium tuberculosis* by cartridge based nucleic acid amplification test (CBNAAT) and anti-tubercular therapy (ATT) was started. The patient was discharged with ICT in-situ and anti-tubercular therapy. Follow-up after 15 days revealed spontaneous closure of BPF and repeat chest radiograph showed complete expansion of lung. Thus, the chest tube was removed, and ATT was continued.

**Image 1:** Chest X-ray of the patient showing ICT insitu (Image 1a). CT thorax of the same patient showed right sided hydropneumothorax with diffuse ground glass opacities (Image 1b)

**Image 2:** The chest X-ray of the patient showed right sided pneumothorax (Image 2a). CT thorax is shown (Image 2b). Post ICT removal, the chest X-ray showed complete lung expansion (Image 2c)

**Image 3:** Chest X-ray of the patient showed a right sided pneumothorax (Image 3a). CT thorax showed a right sided hydropneumothorax with cavitary lesion in superior segment of right lower lobe (Image 3b)
Case 4
A 65-year-old male patient with type II diabetes mellitus on regular treatment presented with complaints of sudden onset of shortness of breath, chest pain, and occasional cough. He was treated at a local hospital previously with a CT severity score of 17 and was on oxygen inhalation and symptomatic treatment. His laboratory investigations showed a CRP of 33.1 mg/dl and d-dimer 1929.64 ng/ml. The total leukocyte count was 23000 cells/mm$^3$. CT thorax was suggestive of left pneumothorax for which intercostal tube was inserted [Image 4(a)]. However, air-leak persisted suggesting a BPF. The patient was treated with intravenous antibiotics, nebulisation, and low molecular weight heparin. Spontaneous closure of BPF occurred after seven days with complete lung expansion. ICT was removed and patient was discharged after 10 days.

Case 5
A 40-year-old female with type II diabetes mellitus was admitted to the high dependency unit (HDU) with chief complaint of cough, dyspnea, and swelling of right lower limb. She was on oxygen therapy at a flow rate of 4 litres/min. Her laboratory parameters revealed a CRP of 200 mg/ml and d-dimer of 2.3 mcg/ml; other parameters were within normal limits. The chest radiograph showed right massive pleural effusion [Image 5(a)]. Intercostal chest tube was given which drained pus. The air-leak however, persisted. The pleural fluid analysis revealed plenty of pus cells and gram-negative bacilli on gram stain; culture showed no growth. The patient was managed with intravenous antibiotics (IV) antibiotics and low molecular weight heparin. Despite of all the antibiotic support her sepsicaemia worsened leading to multiple organ dysfunction and patient finally succumbed.

Case 6
A 27-year-old female, primigravida was admitted with complaints of shortness of breath. The patient had a respiratory rate of 38/min and SpO$_2$ of 88% with 10 litres/min of oxygen at the time of presentation for which she was admitted to the ICU and kept on NIV support. Her CRP was 68 mg/ml and d-dimer was 0.28 mcg/ml. Emergency caesarean section was performed due to decreased foetal movements. The patient developed shock and worsening hypoxemia and diffuse subcutaneous emphysema over the chest following surgery. Chest radiograph was suggestive of left-sided tension pneumothorax and subcutaneous emphysema. The patient was intubated, and ICT was placed on left side for pneumothorax [Image 6(a)] and was later found to have a BPF. She developed acute pancreatitis and septic shock as a complication of pregnancy and eventually succumbed to death.

Discussion
COVID-19 is caused by severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) virus which eventually emerged as a pandemic and has been declared a global health emergency. Over the past two years, there have been many changes in the spectrum of the initial illness and various complications associated with it. Pneumothorax is one such complication that prolongs the hospital stay, increases morbidity and mortality.
Early in the pandemic, the incidence of pneumothorax in COVID-19 patients was reported as 1%.10 Recently, with the second wave, there were increasing reports of pneumothorax in the hospital admitted patients with COVID-19. We encountered few such patients who got complicated with the presence of BPF.

BPF refers to fistulous connection between major, lobar, or segmental bronchus and the pleural space.8 It is mostly recognized as a postoperative complication of pulmonary resection; however, other commonly contributing factors include necrotizing pulmonary infection, persistent spontaneous pneumothorax, chemotherapy, and radiation therapy.8 If untreated mortality ranges from 25% to 71%.10 The probable reason for fistula formation is the weakening of muscles in intercostal space due to continuous ongoing adjacent inflammation and vigorous coughing by the patient that acts as a driving force.

We experienced six such cases where the pneumothorax was associated with BPF in patients with COVID-19 pneumonia with varying natural history and outcomes [Table 1]. In two cases, there was spontaneous closure of BPF. Indigenous method of autologous blood patch was used in only one case, and it proved beneficial. One patient who failed spontaneous closure of BPF complicated with pyopneumothorax was managed by surgery at a higher centre. One patient was found to have a tubercular aetiology for which he is currently on anti-tubercular therapy. Two patients died due to development of sepsis and multiorgan dysfunction. Out of the six cases, three were on NIV support and one required invasive mechanical ventilator support. In all cases, there was a prolonged hospital stay which increased the economic burden and suffering of patients significantly.

### Conclusion

COVID-19 lung complicated with pneumothorax and BPF affects the overall morbidity, and hospital course significantly. It also has an impact on the patient’s economic burden, duration of antibiotic usage, and treatment outcome. The judicious use of steroids, proper ventilation strategy, and close follow-up of high-risk group can minimize the occurrence and complication of BPF. Our case series will enlighten on this emerging entity. It would be a yardstick for further research to prevent occurrence of BPF and proper management of this emerging complication.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and anonymity cannot be guaranteed.

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### Conflicts of interest

There are no conflicts of interest.

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