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Case Report

A Case of Late Bronchopleural Fistula in a Patient of Liver Transplant and Interstitial Lung Disease

Ajoy Krishna Sarkar[1], Arindam Mukherjee[2], Sayan Das[3], Bhaswati Dasgupta Nath[4], Raja Basu[5], Avijit Das[6] and Abhraneel Guha[7]*

[1]Clinical Director of Critical Care and Head of the Unit Pulmonary Medicine, Peerless Hospital and B.K. Roy Research Centre, Kolkata, West Bengal, India

[2]Consultant Respiratory Medicine, Peerless Hospital and B.K. Roy Research Centre, Kolkata, West Bengal, India

[3]Consultant Interventional Radiologist, Peerless Hospital and B.K. Roy Research Centre, Kolkata, West Bengal, India

[4]Consultant Internal Medicine, Peerless Hospital and B.K. Roy Research Centre, Kolkata, West Bengal, India

[5]Consultant Internal Medicine and Intensivist, Peerless Hospital and B.K. Roy Research Centre, Kolkata, West Bengal, India

[6]Consultant Tropical Medicine, Peerless Hospital and B.K. Roy Research Centre, Kolkata, West Bengal, India

[7]Senior Registrar, Internal Medicine, Peerless Hospital and B.K. Roy Research Centre, Kolkata, West Bengal, India

***Corresponding Author:** Abhraneel Guha, Senior Registrar, Internal Medicine, Peerless Hospital and B.K. Roy Research Centre, Kolkata, West Bengal, India.

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Abstract

60-year-old male patient with a background of post liver transplant from living donor 10 years back and also diagnosed interstitial lung disease (ILD) 2 years back presented with a left sided pneumothorax with bronchopleural fistula (BPF) in February this year. He was managed initially in a local hospital with intercostal chest drain. Because of persistent bronchopleural fistula, his left lung remained in a state of partial collapse. He was admitted in our hospital with respiratory distress due to surgical emphysema in addition to pneumothorax. After changing the intercostal drain, surgical emphysema got better but there was no sign of improvement of his bronchopleural fistula. A CT scan of the chest revealed moderate amounts of pneumothorax on left pleural space with ICD seen *in situ*. Three dimensional reconstruction of CT thorax suggested a possible fistula in the left anterior upper segmental bronchus. Localization of BPF was confirmed by inflating a 4F fagoti balloon followed by injecting Iohexol dye in the above subsagement through fibre optic bronchoscopy. Subsequently bronchoscopy guided endobronchial glueing and closure of the BPF was done. Repeat CT scan confirmed closure of the BPF and resolution of the pneumothorax was noted. Patient was discharged in a stable condition.

Keywords: Bronchopleural Fistula; Liver Transplant; Endobronchial Glueing; Fiberoptic Bronchoscopy; Interstitial Lung Disease

Introduction

Solid organ transplant patients are always under different immunosuppressive therapy. Therefore they are susceptible to many bacterial, fungal, viral, parasitic and protozoal infection [1,2]. Bronchopleural fistula occurs in solid organ transplant patients [3]. The causes of this fistula in any post transplant

patients are either as a postoperative complication or secondary to infection. This complication has also been noted in patients of other solid organ transplants due to respiratory infections [3]. Pleural diseases occurring before and after organ transplantation have important implications. Multiple sub pleural bleb due to Staphylococcal pneumonia can cause Spontaneous pneumothorax and also bronchopleural fistula [3]. Bronchopleural fistula are of two types: early and late [4]. Early BPF are due to postoperative complication, late BPF are mainly due to infection. Bronchopleural fistula (BPF) following solid organ transplant (liver and kidney) has been reported before, but is an extremely rare phenomenon [5]. Interstitial lung disease has been associated with bronchopleural fistula [6]. Sherman., *et al*. in his article reported two BPF patients out of the three ILD patients. Exact cause and risk factors were not described though.

We are going to discuss in our case such a rare phenomenon, late bronchopleural fistula with pneumothorax in a patient who underwent liver transplant 10 years ago and developed interstitial lung disease 2 years ago.

Case Report

60 years male with a background of post liver transplantation (on tacrolimus and mycophenolate mofetil) and interstitial lung disease (ILD) had been unwell for the last three months after having a diagnosis of left sided pneumothorax and a bronchopleural fistula. He was discharged recently (one month back) with an intercostal drain (ICD) *in situ* from a local hospital. He had been unwell again and presented two weeks later with increasing dyspnea, cough and chest tightness, necessitating readmission. He was alert, hemodynamically stable but was found hypoxic, saturation was around 88% on room air. Clinical examination revealed reduced breath sounds on the left side. The water seal intercostal drain was showing minimal swing with gross bubbling of intercostal drain. There was also both palpable as well as audible crackles throughout the chest wall suggestive of surgical emphysema.

All necessary investigation were done, which revealed Hb-13.7, TLC-7900, platelets-3.98 lacs, ESR-40, Creatinine-0.78, Na-134, K-4.1, Bilirubin-0.9, Albumin-3.58, Globulin-5, SGPT-45, SGOT-29, Alkaline Phosphatase-125, GGT-97, PT-13.6, INR-1.24. Echocardiography revealed 60% ejection fraction.

HRCT thorax with virtual bronchoscopy revealed moderate amounts of pneumothorax on left pleural space with ICD seen *in situ*. After three dimensional (3D) reconstruction (Image A and B), BPF was suspected in the area of left anterior upper segmental bronchus extending up to the parenchymal surface with a focal scar formation noted likely of previously ruptured parenchymal bullae. Multifocal areas of subpleural tiny cystic changes noted in the anterior and posterior segment of bilateral upper lobe, posterior basal segment of bilateral lower lobe, medial segment of right middle lobe with adjacent subpleural interstitial thickening with ground glass opacity visualized in the posterior segment of bilateral lower lobe, medial segment of right middle lobe, posterior basal segment of bilateral lower lobe suggestive of Interstitial lung disease.

Image A: CT Thorax showing BPF in anterior aspect of left lung.

After admission in our hospital, we changed the intercostal drain by putting a new drain in through different port. Because of persistent left pneumothorax with BPF and as there was a suggestion of BPF in the left upper anterior subsegment, we have decided to localize it more precisely by inflating a 4F Fagoti balloon followed by injecting Iohexol dye in the above subsegment through fiberoptic bronchoscopy. Subsequently bronchoscopy guided glue was injected into the specific sub segment of left upper anterior lobe and closure of the BPF was done. Immediately after the procedure, there was no bubbling of air in the water-seal bag even with cough. Next day, chest x-ray and CT thorax (Image C and D) confirmed closure of the BPF and there was complete resolution of the pneumothorax. Supportive measures and antibiotic courses was given as appropriate. He showed a good recovery and was discharged in stable condition.

Image B: CT Thorax showing BPF (marked by yellow arrow) left upper segment with pneumothorax.

Image D: CT Thorax post procedure showing closure of BPF.

Image C: CT Thorax showing closure of BPF with glue filling (marked by yellow arrow).

Discussion

Bronchopleural fistula (BPF) is a process and not a specific disease [9]. In BPF there's a direct communication between pleural cavity and bronchial tree [9]. CT is also useful in guiding the management procedures for the treatment of BPF [10]. Currently multidetector Computed tomography (CT scan) and advanced applications like virtual bronchoscopy, multiplanar reconstruction, volume rendering images are used as non-invasive procedures in detecting BPF [11]. CT scan with 3D reconstruction can give exact location of BPF and is thus very useful in management of BPF. Chest radiography are valuable in monitoring the efficacy of the therapy [9]. Invasive methods like bronchoscopy, video thoracoscopic exploration or open exploration are also used [11].

Our patient has undergone liver transplant 10 years back and is under immunosuppression. Immunosuppressive patients are at risk of repeated chest infection due to various organism [1,2]. Repeated chest infection could be a cause for bronchopleural fistula [3].

Our patient was having both the conditions, ILD (fibrotic variant of NSIP type) for the last 2 years and the background history of a liver transplant done 10 years ago. As he was asymptomatic for last 8 years, he is less likely to develop BPF due to infections. Thus, ILD seems to be a causative factor.

Low preoperative serum albumin has been labelled as risk factor for late bronchopleural fistula in patients after pneumonectomy for non small cell lung carcinoma in a study by Matsuoka., *et al*

[12]. Research needs to be done to see whether it's a risk factor in patients who underwent liver transplant or for other solid organ transplants. However, in our case, the albumin level was near normal.

Research has mainly been done in BPF patients of lung cancer and lobectomy patients. Jichen., *et al*. in his study tried to analyse the cause of early and late BPF mainly after non small cell lung cancer surgery. In fact, they found chest infection and operative procedure as independent risk factors of early and late BPF [4]. Early BPF was found mainly in aged, late BPF was found in patients who underwent post-operative radiotherapy. They also found that early BPF had higher mortality rates [4].

Persistent BPF has significant comorbidity and mortality for any patient and its correction is mandatory for patients survival. For correction, we have got the following options, open thoracoscopic surgery, video assisted thoracoscopic intervention or bronchoscopic intervention, and endobronchial glueing. Cellulose patch and fibrin glue are also used to repair BPF [13]. Also, Amplatzer device have been used to corrected BPF [14].

Since he had undergone a major surgery (liver transplant and was on tacrolimus and mycophenolate mofetil) previously, open thoracic surgery in such a patient could have been a difficult option and could be even fatal as well. Similar kind of risk also applies for video assisted thoracoscopic correction. So, we opted for a procedure which has got least risk like endobronchial glueing. Endobronchial glue was instilled in our patient after precise localisation of bronchopleural sub-sagement and the nearly 3 months old BPF was closed. His subsequent chest x ray and CT thorax revealed complete expansion of the left lung.

Conclusion

We are reporting this case as our patient who had liver transplant 10 years ago and was diagnosed as ILD about 2 years back, was suffering from left sided pneumothorax with BPF for the last three months. Almost two and half months of indwelling intercostal drain failed to resolve his problem. Surgical correction both for open or video assisted thoracoscopic would have been a difficult procedure and also carries high risk. We could not find any case reporting of BPF in a patient who had liver transplant 10 years back and also suffering from ILD for last 2 years and was successfully corrected by endobronchial glueing through fiberoptic bronchoscopy. Late BPF in a liver transplant patient is a very rare complication to report. The cause in our patient could be due to either ILD or immunosuppression related complication.

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