

Case Report

Giant bronchopleural fistula and empyema in a tuberculosis patient with diabetes mellitus: Vista from a high tuberculosis burden country in Southeast Asia

Budi Yanti^{1,2*}, Saiful Hadi^{1,2}, Fenny Harrika³ and Aamir Shehzad⁴

¹Department of Pulmonology and Respiratory Medicine, School of Medicine, Universitas Syiah Kuala, Banda Aceh, Indonesia; ²Department of Pulmonology and Respiratory Medicine, Dr Zainoel Abidin Teaching Hospital, Banda Aceh, Indonesia; ³Department of Radiology, Dr Zainoel Abidin Teaching Hospital, Banda Aceh, Indonesia; ⁴Disease Diagnostic and Surveillance Laboratory, Bhakkar, Pakistan

*Corresponding author: byantipulmonologis@unsyiah.ac.id

Abstract

Bronchopleural fistula is a pathological tract between the bronchial tree and the pleural space, which can be life-threatening due to tension pneumothorax. It is a rare complication in tuberculosis cases with highly variable in clinical manifestations and persistent air leaks which might lead to complications such as empyema. Herein, we present a tuberculosis and diabetic patient complicated with giant bronchopleural fistula and empyema. A 48-year-old man presented with shortness of breath for two weeks and cough with phlegm for two months. The patient was a smoker with severe Brinkman Index and diabetes. Physical examination revealed hyper resonant percussion and vesicular diminished on the left hemithorax. Laboratory results indicated the patient had anemia, leukocytosis, and hypoalbuminemia. GeneXpert sputum confirmed the presence of *Mycobacterium tuberculosis* and chest X-ray indicated a collapsed left lung. The patient was diagnosed with left secondary spontaneous pneumothorax, pulmonary tuberculosis, and diabetes. The patient was treated with chest tube drainage and anti-tuberculosis drugs. There was no improvement based on serial chest X-ray, and empyema appeared from the chest tube. CT-scan showed tuberculosis lesion, the collapsed of the left lung and fistula in segments 7-8 inferior lobe. Exploratory thoracostomy was performed, in which a giant bronchopleural fistula was detected and then repaired with BioGlue surgical adhesive. Unfortunately, the thoracostomy led to extensive subcutaneous emphysema and was treated by cervical mediastinotomy. The drainage was unable to be removed, and the patient was discharged with Heimlich-type drainage valves on day 28 of treatment. The empyema fluid was cultured and revealed *Staphylococcus haemolyticus*. This case highlights that tuberculosis could cause a bronchopleural fistula and empyema may occur secondary to late diagnosis that needs immediate surgery.

Keywords: Bronchopleural fistula, tuberculosis, empyema, subcutaneous emphysema, *Mycobacterium tuberculosis*



Introduction

Bronchopleural fistula (BPF) is the formation of a pathological tract connecting the bronchial tree to the pleural cavity due to lung tumors, pneumonia, empyema, blunt and penetrating lung

injuries, as well as complications from surgical procedures [1]. Depending on the location, it can be classified into two: central and peripheral BPF. The first is the fistula between the pleura and the tracheobronchial tree, whereas peripheral BPF is between the pleura and the airways distal to the segmental bronchi or lung parenchyma [2].

Tuberculosis (TB), caused by *Mycobacterium tuberculosis* (MTB), is an infectious lung disease with a high prevalence and spreading rate globally [3]. TB can cause various complications such as pneumothorax, empyema, subcutaneous emphysema accompanied by pneumomediastinum, and bronchopleural fistula [4]. Therefore, variation of clinical presentations and complications of TB is important to be studied.

The most common cause of BPF is lung resection, which varies between 4.5–20% and 0.5–1% after pneumonectomy and lobectomy, respectively [1,5]. Its mortality ranges from 18 to 71% [6]. BPF as a complication due to TB is very rare [7]. The exact incidence is difficult to determine since the diagnosis is rarely mentioned in the literature [8]. TB is associated with secondary spontaneous pneumothorax which can develop to BPF and empyema if left untreated [9].

BPF is a severe complication of TB and the clinical condition could be worsened with pneumothorax and pleural empyema [10,11]. Furthermore, the delayed treatment of empyema and pneumothorax leads to lung collapse. TB empyema is an active infection of the pleural cavity, characterized by viscous pus and visceral pleural calcifications. A well-planned and documented conservative medical treatment of TB empyema leads to optimal results. Its management includes chest tube catheter drainage and appropriate therapy for the underlying cause, such as anti-tuberculosis drugs in appropriate doses [12]. This report sought to present a rare case of BPF and empyema caused by MTB infection in a diabetic patient.

Case

A 48-year-old man was admitted to Dr. Zainoel Abidin Teaching Hospital, Banda Aceh, Indonesia with complaints of shortness of breath for two weeks and worsened two days before admission. The patient had been coughing up green phlegm for two months, resulting in chest pain. There was also a complaint of intermittent fever for a month mainly at night, weakness, fatigue, and decreased appetite. Additionally, a weight loss of approximately 15 kg was reported. The patient had smoked up to 32 cigarettes per day since the age of 15-year-old.

On physical examination, blood pressure was 125/82 mmHg, pulse and respiratory rates were 98 and 26 times per minute, respectively and the temperature was 37.8°C. A lung examination revealed an asymmetrical chest wall, where the left stem fremitus was lower than the right, with hyper resonance on the lateral side of the left hemithorax and vesicular disappearance in the left hemithorax.

The laboratory examination showed hemoglobin 8.6 g/dL, hematocrit 27%, leukocytes 30.600/mm³, platelets 406.000/mm³, mean corpuscular volume (MCV) 81 fL, mean corpuscular hemoglobin (MCH) 28 pg, mean corpuscular hemoglobin concentration (MCHC) 32%, red cell distribution width (RDW) 18.6%, eosinophils 1%, basophils 1%, rod neutrophils 0%, segment neutrophils 86%, lymphocytes 5%, monocytes 7%, urea 29 mg/dL, prothrombin time (PT) 14.20 seconds, and activated partial thromboplastin time (APTT) 34.80 seconds. D-dimer was 8.400 ng/mL, hemoglobin A1c (HbA1C) 11.6%, random blood sugar 236 mg/dL, sodium 134 mmol/L, potassium 4.1 mmol/L, chloride 102 mmol/L, albumin 2.2 g/dL, SGOT 25 U/L and SGPT 8 U/L.

The GeneXpert assay on the patient's sputum confirmed the presence of MTB with threshold cycle (Ct<16) and rifampicin resistance was not detected. The sputum culture revealed the growth of *Klebsiella pneumoniae* that was sensitivity to amikacin, cefotaxime, ceftriaxone, gentamicin, and meropenem. The chest X-ray on admission showed fibroinfiltrates in both lungs, and collapsed left lung (**Figure 1A**).

The patient was diagnosed with secondary spontaneous pneumothorax, bacteriologically confirmed pulmonary TB, empyema, anemia, and diabetes mellitus (DM). The left hemothorax was placed on a water-sealed drainage (WSD) and resulted a yellow-green pleural fluid. The empyema fluid was cultured, and *Staphylococcus haemolyticus* was identified. The patient was

treated with intravenous meropenem 1 gram every 8 hours, gentamicin 160 mg daily, intravenous omeprazole 40 mg daily, subcutaneous injection of long-acting insulin 0-0-0-10 IU, anti-tuberculosis drugs based on body weight (rifampicin 450 mg, isoniazid 300 mg, pyrazinamide 1500 mg, and ethambutol 1250 mg daily), paracetamol, codeine, and gabapentin. The anemia was corrected by a transfusion of two packed red cell (PRC).

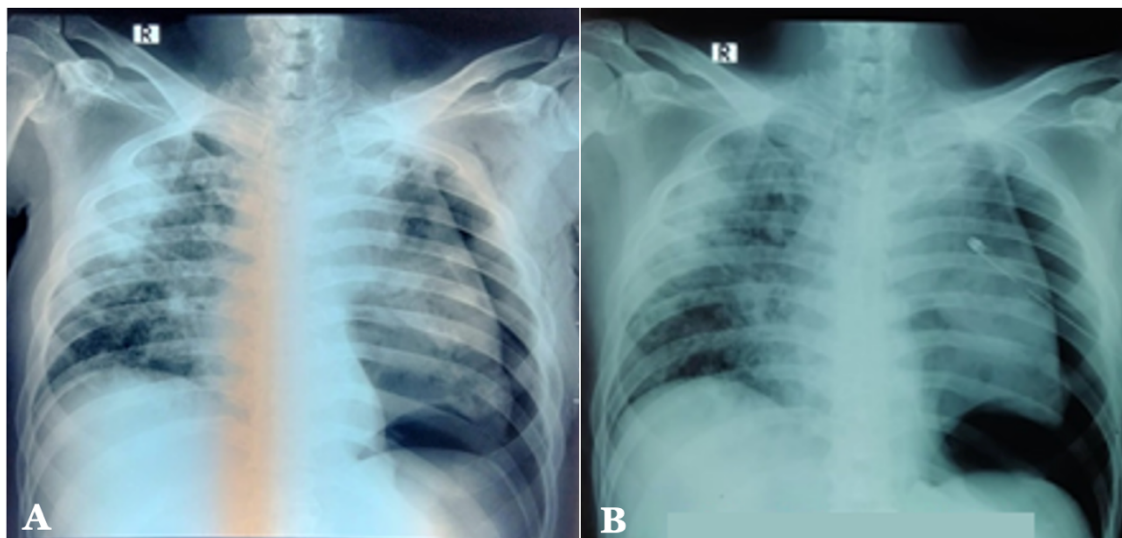


Figure 1. (A) Chest X-ray on admission and (B) chest X-ray after two weeks of hospitalization.

After two weeks of hospitalization, there was no lung expansion (**Figure 1B**). Air bubbles and empyema formation were discovered in the WSD. Subsequently, the patient underwent a chest computerized tomography (CT)-scan, which showed extensive fibrotic lesions in the upper right lung field with multiple cavities. Fibrosis was also present at the apex of left lung, which collapsed with a firmly and laterally hypodense area. Also, a fistula was suspected in segments 7–8 of the inferior lobe of left lung (**Figure 2**).

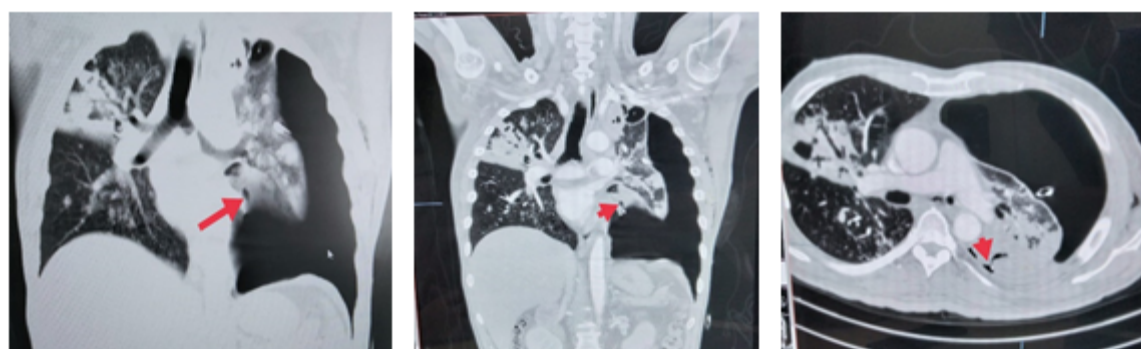


Figure 2. Computerized tomography scan of the patient. The red arrow indicates a bronchopleural fistula.

The exploratory thoracotomy was conducted to repair the bronchopleural fistula (**Figure 3**). The intraoperative course discovered bullae in the inferior lung lobe and a fistula with a size of 0.5 cm x 0.5 cm and a diameter of 1 cm, which was sutured tightly and closed using BioGlue surgical adhesive. The WSD tube was retained for drainage and subsequent management evaluation. After the thoracotomy, there were complaints of severe shortness of breath and subcutis emphysema in the facial region, thoracoabdominal, both sides of brachii and antebrachii.

Subcutaneous emphysema was treated with cervical mediastinoscopy. This was retained on the patient for 28 days and showed clinical and laboratory improvement, after which the patient was discharged with a thoracic drain with a Heimlich valve.

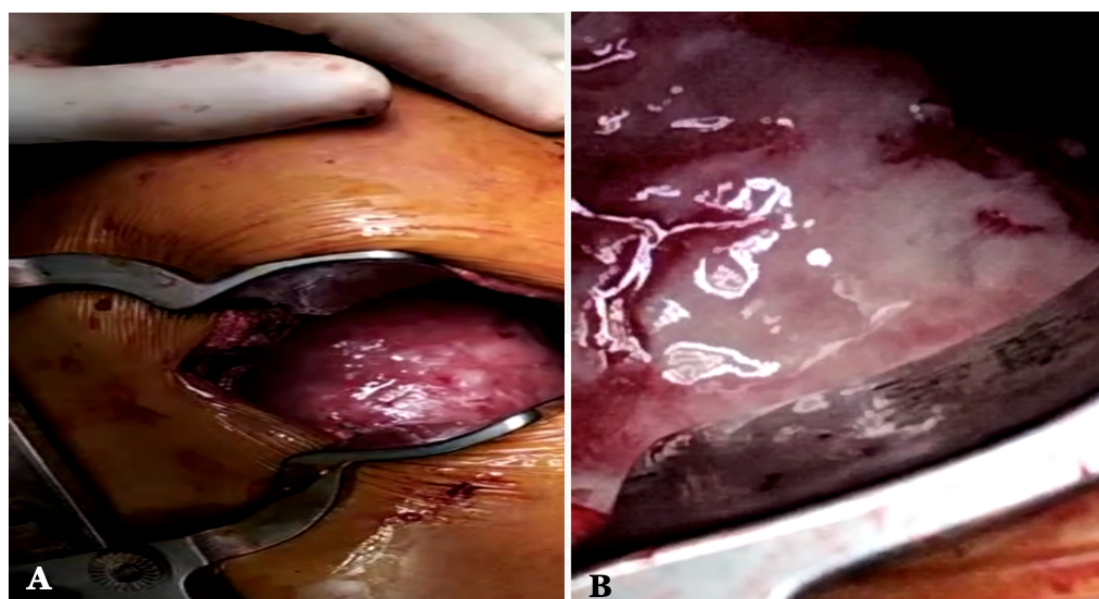


Figure 3. Exploratory thoracotomy (A) and bullae in the inferior lung lobe (B).

Discussion

The clinical manifestations of patients with BPF depend on the size and duration of infection. The patient usually complains of fever, cough with mucoserous or purulent phlegm, night sweats, and chills [13]. The productive cough worsens when the patient lies on the opposite side of the fistula's hemithorax. Furthermore, a typical sign of this complication is the massive fluid production in WSD within a few days of treatment with symptoms of pleural leakage the bubbles in drainage [14].

We presented a diabetic man with DM and worsening shortness of breath. The patient had TB with a pneumothorax that did not improve after chest tube drainage. Currently, DM is one of the most common comorbid that enhances the susceptibility to MTB infection. DM with its macro and microvascular complications and smoking habits increase the progressivity of TB [15].

The CT-scan characteristic of BPF generally shows a passage from the bronchi or lung parenchyma to the pleural space clearly and thus underlies the definitive diagnosis of BPF. CT-scan could detect 50% to 76% of BPF cases with thin slices and multisectional projection images [16]. The definitive diagnosis of the fistula was established with precise imaging in the branching of the bronchi or lung parenchyma penetrating the pleural cavity [17]. Some studies showed that the thoracic CT-scan sensitivity was 50% in demonstrating the presence of peripheral BPF [4,17]. A study found that the chest CT-scan is able to identify direct or indirect signs in 86 and 100% of central and peripheral BPF patients, respectively [19].

Management of BPF requires a long treatment time, but the first step is to prevent life-threatening clinical deterioration such as sepsis, tension pneumothorax, and respiratory failure [20]. Protecting the contralateral lung from aspiration of pleural fluid is essential to reduce the risk of pneumonia and respiratory failure. Therefore, installing a WSD to ensure drainage of the pleural cavity is necessary. Depending on the antibiotic suitable for the pleural fluid culture results, broad-spectrum antibiotic therapy sensitive to Gram-positive, Gram-negative, and anaerobic microorganisms needs to be administered [21].

Generally, the BPF management aims to re-perform a thoracotomy to treat fistula formation. A study reported conservative management of post-resection BPF in 17 case and successful closure was achieved in 16 cases [22]. The various bronchoscopy techniques used to treat small BPFs ranging from 0.8 to 1.0 mm include sealants, fibrin glue, coils, and endobronchial silicone or metal stents. Dutau et al. used a self-expanding metal stent in large

fistulas (>6 mm) and reported a postoperative improvement in the patients' breathing [23]. Conservative management includes WSD insertion and treatment of the primary etiology with anti-TB drugs at appropriate doses. The treatment of empyema due to TB could be maximized by conducting a concentration test between the serum anti-TB drug level and the pleural fluid [11]. Furthermore, inflammation of the pleural membranes due to acute TB infection is rarely diagnosed. Further measures in the form of pleuroscopy and decortication need to be considered when empyema is discovered in TB cases that do not improve after conservative treatment evaluation [23]. Decortication is an extensive surgery and alternative therapy modality to reduce parietal pleural thickness, increasing lung expansion capacity [24]. In this case, surgery is preferably performed after sterilization of the empyema and requires at least 6 weeks of strictly controlled anti-TB treatment.

Conclusions

TB could lead to the formation of BPF and empyema could occur due to late diagnosis. This complication causes high morbidity and mortality in pulmonary TB patients. Therefore, surgery is needed to eliminate the source of infection, improve treatment responses, repair the fistula and reduce mortality.

Declarations

Ethics approval

Not applicable for case report.

Patient's consent

The patient provided informed consent to be included published as case report.

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Conflict of interest

Authors declare no conflict of interest.

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