Tuberculosis is responsible for considerable morbidity and mortality worldwide. The presentation and complications of pulmonary tuberculosis are quite diverse. Aktogu et al investigated 5,480 patients with pulmonary and pleural tuberculosis, and about 1.5% had pneumothorax. However, recurrent bilateral pneumothoraces as a complication of miliary tuberculosis is extremely rare. We report a case of bilateral, simultaneous pneumothorax as a complication of miliary tuberculosis, and pneumothoraces that recurred and which were treated effectively by surgery.

**Case Report**

A 22-year-old woman from Thailand was admitted to the hospital because of nonproductive cough and fever for 3 days. Two months before admission, she had received a health examination, and her chest radiograph was normal. Two weeks before admission, she started to have poor appetite and general malaise. Progressive dyspnea and chest tightness developed over the following week. Three days before admission, dry cough, persistent fever and chills developed. Apart from a high
temperature (38.2°C), physical examination on admission produced normal results. Chest radiography showed diffuse, interstitial shadows with miliary nodules throughout both lung fields (Figure 1A). White blood cell count was 3700/mm³, with a differential of 84.8% polymorphonuclear leukocytes and 12.2% lymphocytes. Hemoglobin was 12.9 mg/dL and platelet count was 211,000/mm³. Serum biochemistry studies produced normal results. Sputum smears produced negative results for acid-fast bacilli on three consecutive occasions. Cultures of sputum, urine and blood for pathogens produced negative results. The serology test for human immunodeficiency virus also produced a negative result. The patient was treated on admission for a presumed diagnosis of miliary tuberculosis with isoniazid (INH), rifampin (RIF), ethambutol (EMB), and levofloxacin. The dyspnea gradually improved, but intermittent high fever persisted.

On the 27th hospital day, the patient developed sudden onset of dyspnea and chest pain, and chest radiography showed bilateral pneumothorax (Figure 1B). Chest tube thoracostomy was performed, with almost complete expansion of both lungs within 24 hours. On the 30th hospital day, the patient experienced dyspnea and pain on the right side of the chest and chest radiography revealed right pneumothorax. Contrast-enhanced computed tomography of the chest revealed fine nodules uniformly distributed throughout both lungs and a right pneumothorax (Figure 2). Chest tube drainage was instituted and video-assisted thoracoscopic (VATS) lung biopsy of the right lung was performed. During operation, several blebs

Figure 1. (A) Chest radiography, posteroanterior (PA) view, on admission shows diffuse, interstitial shadows with miliary nodules throughout both lung fields. (B) Chest radiography, PA view, obtained right after sudden onset of dyspnea on the 27th hospital day shows bilateral pneumothorax.

Figure 2. Contrast-enhanced computed tomography of the chest demonstrates diffuse and widely dispersed small nodules uniformly distributed throughout both lungs and a right pneumothorax.
on the lung surface were found (Figure 3); they were grasped with ring forceps and excised with a 45-mm endoscopic stapler. Pathology showed bleb formation with granulomatous interstitial pneumonia and miliary bronchiolocentric granulomas; acid-fast positive bacilli were found via the Kinyoun method. Lung tissue culture for *Mycobacterium tuberculosis* produced positive results. Miliary tuberculosis was confirmed. However, left tension pneumothorax occurred the next day, and chest tube drainage was immediately performed. After receiving antituberculous treatment for 2 months, her fever gradually subsided. In the following 2 months, bilateral pneumothorax recurred twice and chemical pleurodesis with minocycline was performed on both sides but air leakage persisted. VATS pleurodesis was performed on both sides in the 16th week of hospitalization. VATS pleurodesis was performed in a standard fashion under general anesthesia, using intubation with a double-lumen endotracheal tube. The patient was placed in a lateral decubitus position, and the ipsilateral lung was deflated. A 10-mm 30° telescope was first inserted through the previous chest tube wound to examine the pleural cavity. Two 15-mm skin incisions were made at the third or fourth intercostal space, anterior and posterior axillary line. Light pleural adhesions were freed by electrocautery and blebs on the lung surface were excised. Twenty milliliters of 2% lidocaine hydrochloride (400 mg) followed by a solution of 20 mL of normal saline containing 300 mg of minocycline were instilled into the pleural cavity. A chest tube was placed in the apex through one of the insertion wounds. Then we repeated the above procedure on the other side. Both chest tubes were removed in the 17th week of hospitalization, without recurrence of pneumothorax on either side. After 4 months in the hospital, the patient was discharged on a regimen of INH, RIF and EMB. There was no further recurrence of pneumothorax during the 3 months of follow-up.

**Discussion**

Pneumothorax is a well known complication occurring in cavitary tuberculosis. However, its occurrence as a complication of miliary tuberculosis is relatively uncommon. Up to now, there have only been 19 cases of miliary tuberculosis complicated with pneumothorax reported in the English literature. Of the 19 reported cases, 11 had unilateral pneumothorax and eight were bilateral. In most of the cases (17/19), the pneumothorax was on the left side or started on the left side before pneumothorax developed on the right side. In all of the bilateral cases, the second pneumothorax developed after admission while the patients were being treated with antituberculous therapy. They received either chest tube thoracostomy or needle aspiration for their pneumothoraces and none of them were treated by surgery. Only three reported cases died. In our case, bilateral pneumothoraces recurred three times during the treatment period. To our knowledge, our patient is the first case who was treated effectively by a combination of antituberculous therapy and surgery.

The pathogenesis of pneumothorax in cavitary tuberculosis can be readily explained by the rupture of the cavities into the pleural space. Although the exact pathogenesis of this complication is not clear in miliary tuberculosis, there are several possible mechanisms. One mechanism might be the formation of small areas of confluent subpleural miliary nodules that undergo caseation.
and necrosis with subsequent rupture of the caseation into free pleural space.2 Alternatively, a bullous lesion might form near the miliary tubercles which rupture to produce a pneumothorax.3,6 In our case, lung biopsy pathology showed bleb formation with granulomatous inflammation, which provides strong evidence that confluent subpleural miliary nodules may form small blebs which subsequent rupture into free pleural space and lead to pneumothorax. Another possible mechanism explaining the pneumothoraces in miliary tuberculosis is interstitial emphysema. This mechanism was described by Peiken et al.2 Miliary nodules are believed to lead to an increased intra-alveolar pressure in patients coughing violently, performing the Valsalva maneuver. The increased pressure leads to air tearing under the alveolar septa and dissecting its way up the peribronchial and perivascular spaces to the mediastinum and mediastinal emphysema may develop.2,3 Initial pneumomediastinum with air leakage through the mediastinal pleura may lead to the development of bilateral pneumothorax. In most reported cases, however, pneumothorax occurred without evident pneumomediastinum.5,7 In our case, the pneumothorax was bilateral but there was no evidence of pneumomediastinum.

By definition, patients with secondary spontaneous pneumothorax have underlying lung disease and they usually have limited cardiopulmonary reserve. The occurrence of pneumothorax can be life-threatening. Therefore, consensus among experts is that aggressive treatment is warranted in patients with secondary spontaneous pneumothorax. After tube thoracostomy to re-expand the lung, further treatment depends on the patient’s general condition and whether prior pleurodesis has been performed. If a patient’s medical condition is good and he/she can tolerate selective one-lung ventilation and general anesthesia, VATS is recommended.10 VATS for secondary spontaneous pneumothorax has been shown to be associated with a higher morbidity, unlike VATS for primary spontaneous pneumothorax.11 Therefore, careful patient selection and clinical judgment are important factors for ensuring optimal outcome. Our patient was a young and previously healthy woman who had a fair cardiopulmonary reserve. Therefore, we judged that VATS pleurodesis would be a safe and effective treatment for her recurrent pneumothorax.

Doddoli et al reported on a series of 39 patients who were treated with VATS for recurrent primary spontaneous pneumothorax after previous talc pleurodesis.12 The VATS procedure was successfully achieved in 27 patients (69%) and postoperative morbidity was limited to pleural complications in the VATS group (n = 6, 22%). Only one patient treated with VATS developed partial recurrent pneumothorax at 12 months with a favorable outcome without further surgery. Although we performed chemical pleurodesis for this patient, we think that VATS is a feasible and effective treatment for her recurrent spontaneous pneumothorax.

In 13 of the 19 cases reported, pneumothorax developed during admission while the patients were on antituberculous therapy.7 In our case, pneumothorax developed on the 27th day of treatment. Our experience with this patient highlights the fact that pneumothorax should be suspected in an adult with miliary tuberculosis who suddenly develops acute respiratory distress, even though this is a rare complication. It can be seen both in early and late periods of treatment. Recurrent pneumothorax can be managed, apart from medical therapy of miliary tuberculosis, with tube thoracostomy and surgical intervention.

References


