Case Report





# Silicotuberculosis with Esophagobronchial Fistula and Broncholithiasis

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## Abstract

A 69-year-old woman was admitted to hospital 4 times from November 2007 to June 2009. The patient had silicosis complicated by broncholithiasis, esophagobronchial fistula, and relapsed tuberculosis. She had worked as a stone crusher for 3 years and was exposed to a large amount of quartz dust. Barium esophagography, gastroesophageal endoscopy, and biopsy suggested esophageal-related chronic inflammation and ulcer, which probably caused the repeated esophagobronchial fistulas observed. Bronchoscopy revealed a free broncholithiasis in the left main bronchus. The patient was readmitted a fourth time, for the relapse of silicotuberculosis. After 9 months of antituberculous therapy, she was doing well until the recent last follow-up visit.

**Keywords:** Silicosis; Esophagus; Fistula; Tuberculosis; Lung diseases

## Introduction

Herein, we present a case of silicosis complicated by broncholithiasis, esophagobronchial fistula, and relapsed tuberculosis. This case demonstrates one of the unusual and complex complications of silicosis. We believe the silicosis complications are interrelated, which differ from previous case reports.[1-4]

## Case Report

A 69-year-old woman was admitted to hospital in November 9, 2007 with acute exacerbation of chronic cough and dysphagia with solids and liquids for 2 weeks. She did not have hemoptysis, fever, chills, chest pain, or weight loss. When she was 9 years old, she had worked as a stone crusher for 3 years and was exposed to a large amount of quartz dust for not using any respiratory protective equipment. In 1969, she was diagnosed with silicosis and tuberculosis. She was treated for tuberculosis and was stable. She had no further exposure to dust or other toxic substances, thereafter. On follow-up, her condition had no change. The patient was a life-long nonsmoker and consumed no alcohol. Nor did she receive any medications.

On physical examination, the patient looked well, with no palpable peripheral lymph nodes. Occasional wheezing was heard on both lung fields. Laboratory data, including blood tests and serum electrolytes, liver enzymes, and bilirubin, as well as renal function were normal. Skin test for tuberculosis was negative. Three consecutive samples of sputum were negative for acid-fast bacilli by staining and culture. Chest x-ray and computed tomography

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(CT) of the chest demonstrated eggshell calcification of the hilar and mediastinal lymph nodes in the paratracheal, subcarinal, and aorticopulmonary window regions. Bilateral multiple pulmonary nodules were also visible. The lower lobe of the left lung showed a ground-glass, dense shadow. Barium esophagography revealed two fistulas between the middle esophagus and the left main bronchus, immediately tracking into the left bronchial tree (Fig 1). The diagnosis was silicosis with esophagobronchial fistula and aspiration pneumonitis. The patient received antibiotics. An esophageal stent was placed on December 14, 2007. The symptoms improved.

The symptoms however recurred within three days. Gastroesophageal endoscopy demonstrated a small ulcerated lesion above the metallic stent, with a small diverticulum 18 cm from the upper incisor; the esophageal mucosa was inflamed and friable. Biopsy revealed chronic inflammation and an ulcer without granulomas, malignancy, or organisms (Fig 2). Another esophageal stent was placed on December 20, 2007. The symptoms improved and the patient was discharged from hospital.

After two months, the patient was admitted to another hospital because of severe cough exacerbated by swallowing liquids and solid foods. Gastroesophageal endoscopy revealed a new small mid-esophageal ulcer. Other examinations, including tests for tumor markers, were normal. A tuberculin skin test gave negative results. Over the next few days, the patient reported expectoration of small grayish coral-like calculi with a stony consistency, measuring 5–10 mm in diameter, as well as episodes of coughing. Bronchoscopic examination showed a free broncholithiasis in the left main bronchus, mucosal thickening, and stenosis with no significant airway obstruction of the main bronchus. The broncholithiasis was extracted with no bleeding. Biopsy of the bronchus revealed chronic inflammation, with no evidence of malignant tumor cells or granulomas. Sputum and bronchoalveolar lavage analyses did not reveal *Mycobacterium tuberculosis*, fungi, or malignant cells. The diagnosis was silicosis with broncholithiasis and esophagobronchial fistula.

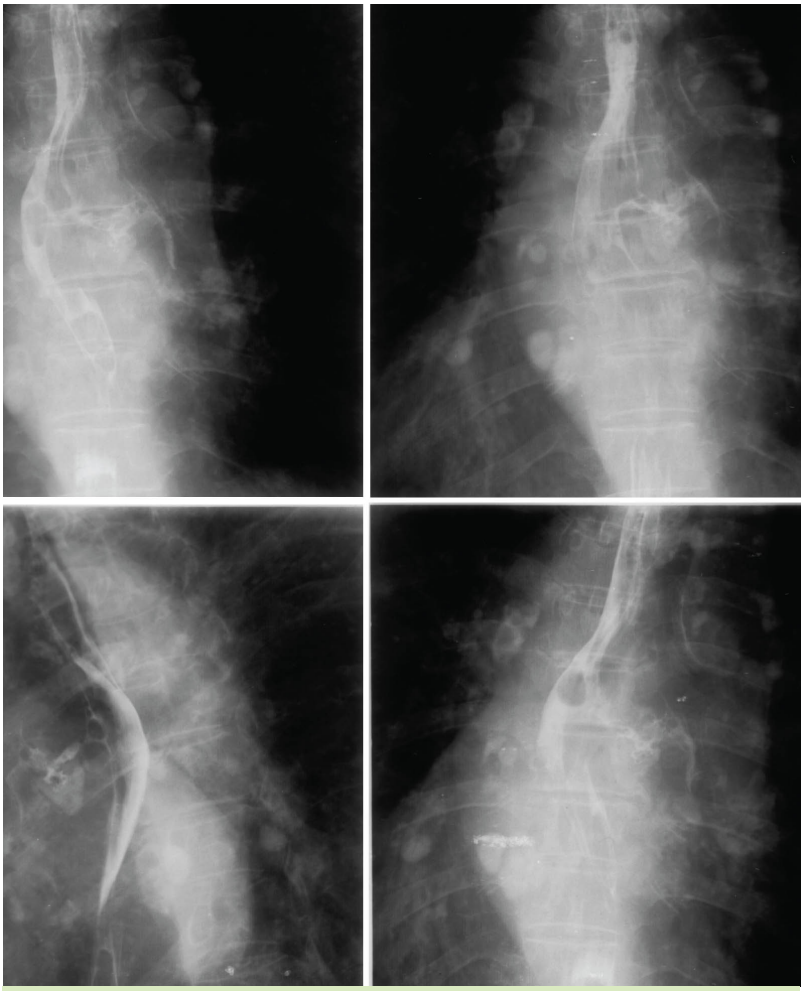


**Figure 1:** Barium esophagography of 2 fistula tracts between the esophagus and bronchus in the patient

The patient continued to have frequent complications and persistent fistula probably related to stasis in the esophagus. Because of her frail condition, we decided to initiate a conservative operative approach and inserted a gastrostomy tube.

Two samples of the expectorated ma-

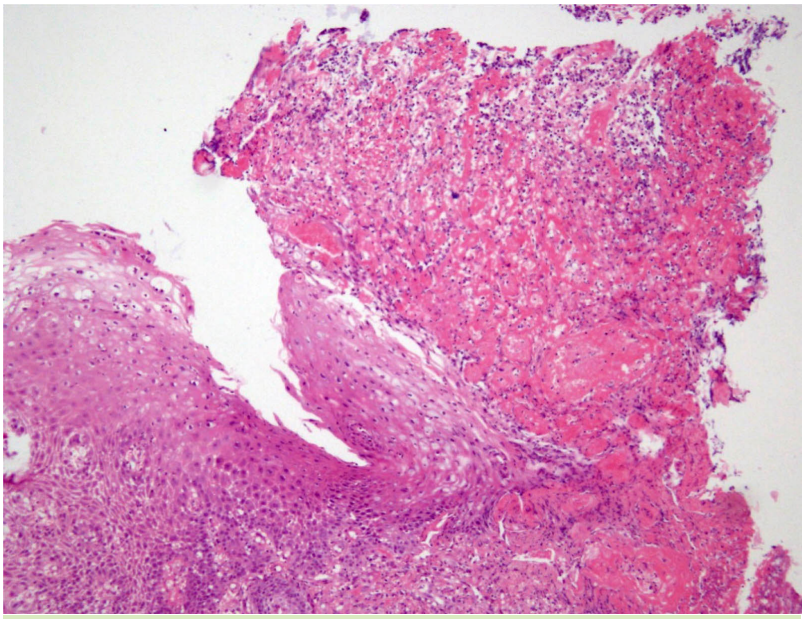


**Figure 2:** Histological examination of esophageal biopsy showing moderate chronic inflammation and ulceration of esophageal squamous epithelium

terials were analyzed by electron microscopy and energy-dispersive spectrometry (EDS), with a Phenom ProX system (Phenom-world Co, the Netherlands), operating at 15 kV. Without any coating on the sample, with SDD high resolution and a high-sensitivity EDS detector, semiquantitative chemical analyses were performed. Mineralogy analysis revealed the broncholithiasis was composed of calcium and phosphate, with no crystals of silica or silicate (Fig 3).

After 16 months, in June 2009, the patient was readmitted because of cough, sputum, dyspnea, fever, and night sweating for 10 days. The admission laboratory data included a white blood cell count of $6.65\times10^3/\mu L$ (reference range $4.0–10.0\times10^3/\mu L$), hemoglobin level of 11.4 g/dL (11.0–15.0 g/dL), high-sensitivity C-reactive protein level of 26.38 mg/L (0–3 mg/L), serum albumin level of 4.32 g/dL (35–55 g/dL), serum protein level 7.0 g/dL, erythrocyte sedimentation rate 34.9 mm/h (0–20 mm/h). A skin test for purified protein derivative (5 TU) was strongly positive, with an induration of 25 mm with ulceration. The concentrated sputum test result was positive. The diagnosis was silicotuberculosis relapse. CT revealed an esophageal stent shadow, esophageal wall thickening, multiple small nodule spots, patchy shadows, and mediastinal and lung door multiple calcified lymph nodes (Fig 4). Antituberculous treatment with 450 mg rifapentine, twice a week, 300 mg inhaled isoniazid, once a day, and 750 mg pyrazinamide, once a day, was started. After nine months of antituberculous treatment, the patient did well. We continued to follow the patient until the time we wrote this report; she had no change (Fig 4).

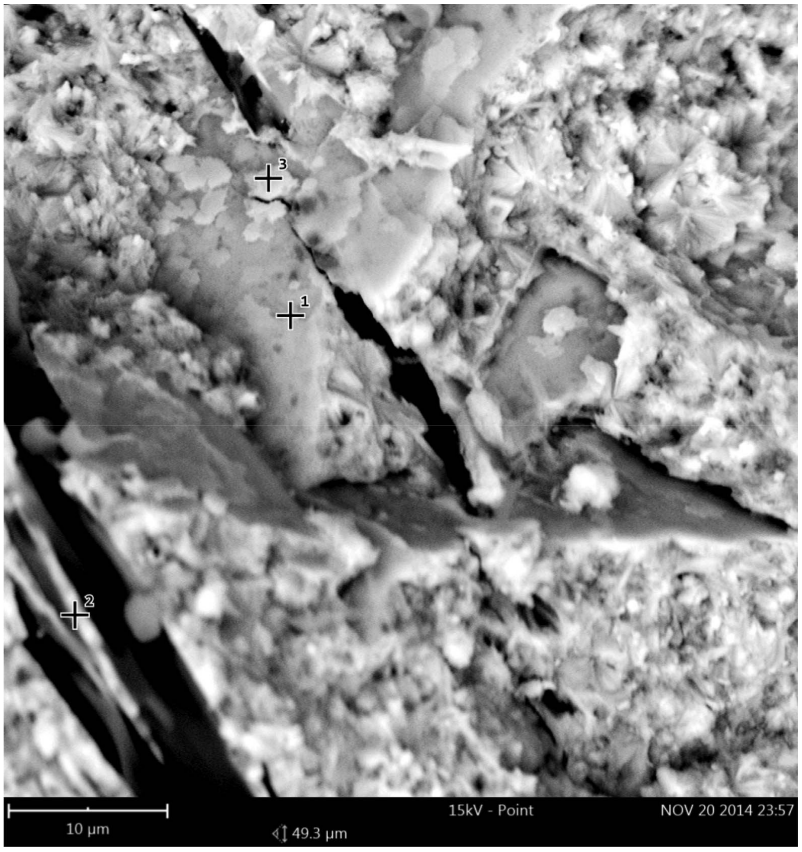
## Discussion

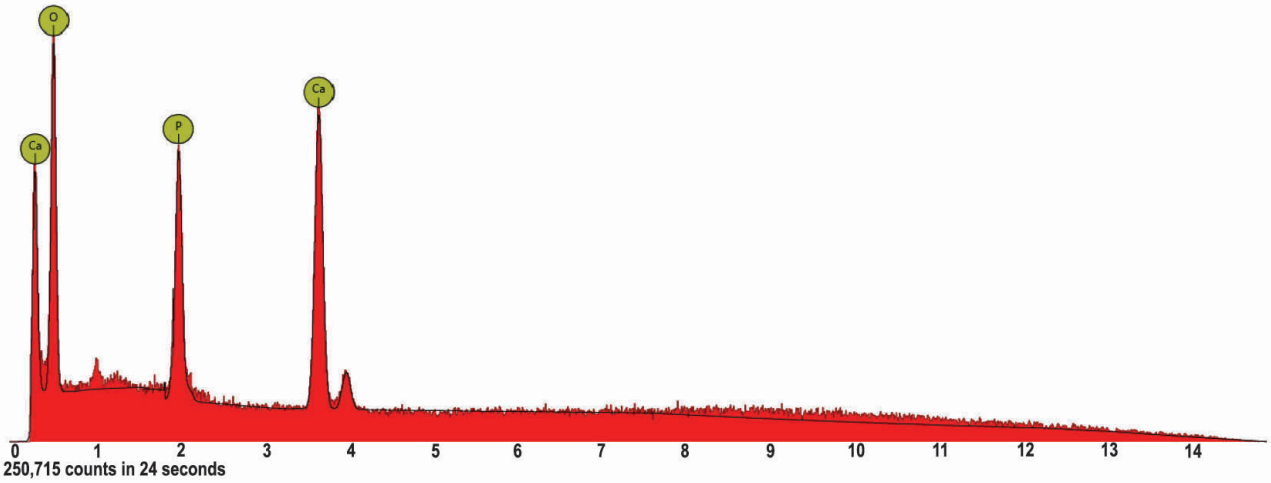
Most esophagobronchial fistulas in adults result from a locally advanced esophageal or bronchogenic malignancy. Benign esophagobronchial fistulas are usually acquired secondary to traumatic or prolonged intubation of the trachea or esophagus,[5] and result in mediastinal inflammation. Silicosis-related lymphadenopathy in the posterior mediastinum with calcification causes a tiny traction diverticulum of the esophagus. We considered that the pressure from the lymph node had caused the esophageal torus lesion and that obstruction of the bloodstream had caused the ulcer. Ingested sharp silica fragments may cause local repeated injuries during their passage down the esophagus and be buried in the mucosa, stimulating proliferation by providing anchorage.[6] From the barium esophagography, gastroesophageal endoscopy examination, and biopsy, the patient may have esophageal-related chronic inflammation, diverticulitis and ulcer, which may be causes of repeated esophagobronchial fistulas. The frequent complications and persistent fistula were probably related to stasis in the esophagus.

Silicosis is frequently complicated by tuberculosis.[7] Several studies have reported that mediastinal granulomatous inflammation caused by tuberculosis could account for 14% to 67% of benign esophagobronchial fistulas, according to the prevalence of tuberculosis in the study population.[8] An esophagobronchial fistula is an unusual manifestation of tuberculosis, and its correct diagnosis may sometimes be difficult because of an insidious and nonspecific clinical course.

Although the tuberculosis-related examination failed to find activities, tuberculosis could not be ruled out. More than one year later, the patient showed symptoms of tuberculosis, eventually establishing the diagnosis, which was successfully treated with antituberculous chemotherapy. We believe that the initial tuberculosis could not be excluded.

Traditionally, the treatment of esophagobronchial fistula has relied on surgical repair. In recent years, medical manage-




| Element Number | Element Symbol | Element Name | Confidence | Concentration | Error |
|---|---|---|---|---|---|
| 20 | Ca | Calcium | 100.0 | 10.1 | 0.9 |
| 8 | O | Oxygen | 100.0 | 82.6 | 1.1 |
| 15 | P | Phosphorus | 100.0 | 7.2 | 1.1 |

**Figure 3:** Electron microscopy and energy-dispersive spectrometry of broncholithiasis sample

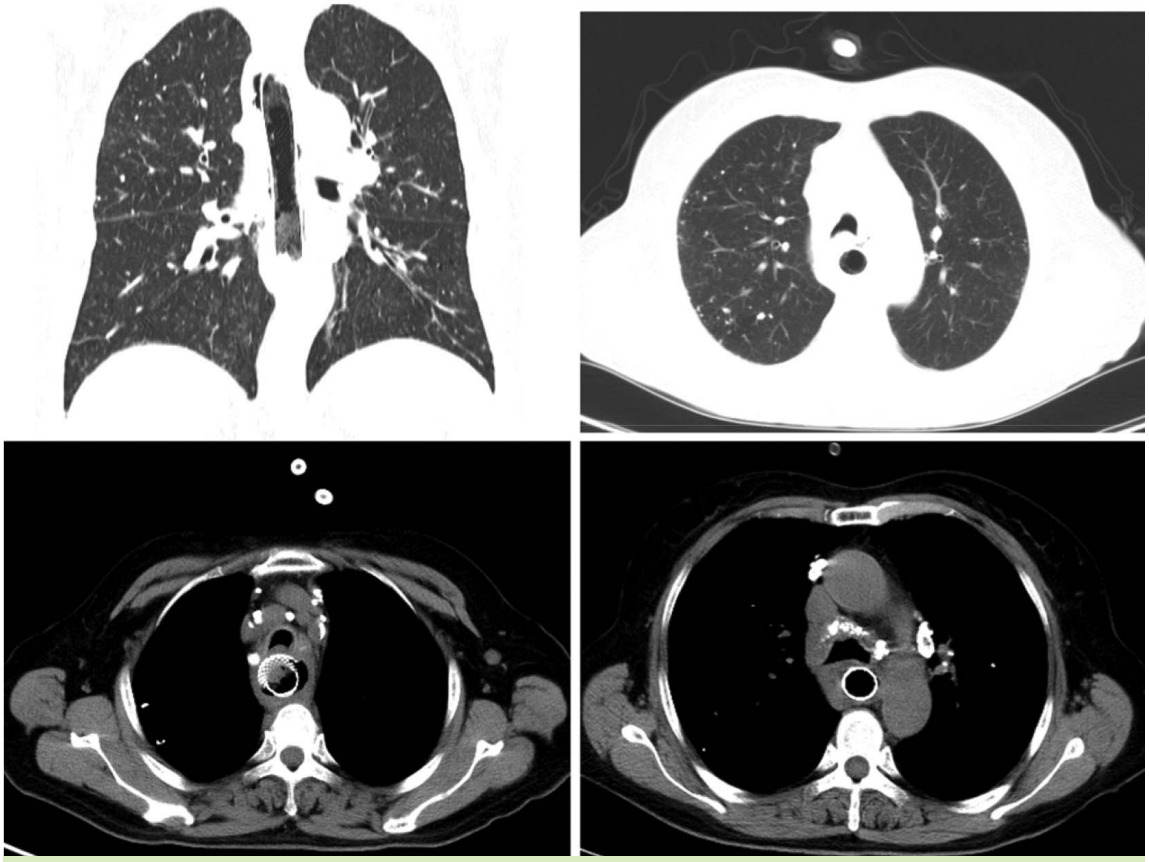


**Figure 4:** CT scan of esophageal stent shadow, esophageal wall thickening, multiple small nodules, patchy shadows, and mediastinal and lung door multiple calcified lymph nodes.

ment with antitubercular therapy for patients with tubercular esophagobronchial fistula has been successful.[9] Other treatments are placement of self-expanding metallic stents, percutaneous gastrostomy or silicon esophageal prostheses.[10] Metallic stents are superior to other methods.[11] For our patient, two esophageal stents were placed for the management of the fistulas but they failed to prevent repeated fistula-related esophageal inflammation. The patient has had a percutaneous gastrostomy to date. The treatment of esophagobronchial fistula depends on the severity of symptoms, the location of the fistula, and the general condition of the patient.

Broncholithiasis most commonly arises from erosion and extrusion of chronically inflamed calcified contiguous mediastinal or intrapulmonary lymph nodes into the tracheobronchial tree. Previous infection with tuberculosis and histoplasmosis accounts for most cases of broncholith formation, but silicosis is the known noninfectious cause. Silicotic lymph nodes with eggshell calcification can erode a bronchial wall. Sartorelli was the first to describe broncholithiasis in patients with silicosis in 1957;[12] thereafter, a few cases with silicosis were reported.[2,13] Mineralogy analysis of our patient revealed that the expelled substance was composed of calcium and phosphorus, but not crystals of silica or silicate. Energy-dispersive microanalysis of the nodal tissue revealed a sharp peak for silicon.[14] The broncholithiasis was perhaps associated with the previous tuberculosis. Studies have shown that treatment

of broncholithiasis is restricted to clinical follow-up. Some invasive procedures may be necessary, depending on the location, size and associated complications. Generalizing the prognosis is usually favorable.

In our case, the patient's occupational history and radiological features of the disease helped us with the diagnosis of silicosis; however, complications of silicosis are difficult to understand. Attempting to put all these features into one package, we think silicosis of mediastinal lymph nodes was caused by esophagobronchial fistula and broncholithiasis initially. As the disease progressed, relapse of silicotuberculosis emerged. Here, we should emphasize that the complications of silicosis should not be overlooked.

**Conflicts of Interest:** None declared.

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