

中文題目：擬似腫瘤的支氣管結石症導致致命性大咳血

英文題目：Broncholithiasis mimicking malignancy presented with life-threatening hemoptysis

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**Background:**

Broncholithiasis is a rare disease and defined as calcified or ossified materials within the tracheobronchial tree<sup>1</sup>. The prevalence is 0.1-0.2% in the patients with respiratory disease<sup>2</sup>. Although the presentation may be asymptomatic or nonspecific, it may cause fatal hemoptysis. The strategy for massive hemoptysis may be different between broncholithiasis and malignancy based on the pathophysiology.

**Method:**

We reported a patient with broncholithiasis which presented like malignancy at chest computed tomography and caused life-threatening hemoptysis.

**Result:**

Mr. Ye was a 71-year-old retired iron factory employee presented with cardiac arrest following massive hemoptysis. He smoked 55 pack-year and had a 2-month history of cough with little bloody tinge without fever, dirty sputum, shortness of breath, or chest pain. Chest X ray showed mass lesion beside the right heart border (Figure 1). Computed tomography (CT) revealed calcified mass with lung collapse over right middle lobe (Figure 2), and enlargement of lymph nodes with calcification at mediastinum and hilum (Figure 3 & 4). Malignancy or granulation tissue was suspected. Bronchoscopy showed airway distortion, inflamed mucosa, and easy blood oozing when touched. Endobronchial ultrasound-guided bronchoscopic biopsy was done. He was quite well after the examination.

One week later, suddenly massive hemoptysis occurred and he was collapse following blood suffocation. Cardiopulmonary resuscitation (CPR) was done immediately and spontaneous circulation recovered after 60-minutes CPR. Although the bleeding was stopped after the medication and blood transfusion, he was died from the progression of aspiration pneumonia and unstable hemodynamic status.

The pathologic report of the mass showed neutrophil, histiocyte, and lymphocyte infiltration without malignant cell identification. According to the history, imaging, and pathologic finding, peribronchial broncholithiasis complicated with life-threatening hemoptysis was the final diagnosis.

### ***Discussion:***

Broncholithiasis is a rare disease and may be misdiagnosed as malignancy on imaging. However, the strategy for massive hemoptysis treatment may be different between broncholithiasis and malignancy based on the pathophysiology.

The definition of broncholithiasis is presence of calcified or ossified materials within the tracheobronchial tree<sup>1</sup>. The prevalence is 0.1-0.2% in patients with respiratory disease and 0.8% in hemoptysis population<sup>2</sup>. Necrotizing granulomatous diseases with mediastinal lymphadenitis, such as tuberculosis and histoplasmosis, are frequently related to the cause of broncholithiasis. But, the exact mechanism of the calcified materials eroding into airway is elusive. The manifestations varied from asymptomatic to life-threatening, such as massive bleeding or air trapping due to acute airway obstruction. Persistent cough, bloody sputum or hemoptysis, wheezing and chest pain are common symptoms. Lithoptysis is a unique but rare feature with prevalence of 6-26%<sup>1</sup> in patients with broncholithiasis.

CT and bronchoscopy are essential for the diagnosis and classification of the broncholithiasis. Three subtypes include endobronchial type, transbronchial type, and peribronchial type<sup>1</sup>. More stenosis or ectasis of the distal bronchial is found by CT in the patients with endobronchial broncholiths. In transbronchial ones, more bronchial twist and obstructive pulmonary emphysema are found by CT, while thickening and edema of mucosa, bleeding, and lots of sputum are major findings by bronchoscopy. In peribronchial ones, obstructive inflammatory lesion is found by CT, while granuloma, luminal stenosis and occlusion are found by bronchoscopy<sup>3</sup>. Sometimes, the imaging on CT mimics malignancy, and even the positron emission tomographic scan with <sup>18</sup>F-FDG reveals increased uptake, which may suggest malignancy and lead to misdiagnosis<sup>4,5</sup>.

Usually, conservative management with observation is indicated for patients with asymptomatic broncholithiasis. Bronchoscopic removal is the first line treatment for endobronchial and transbronchial broncholithiasis. Surgery may be needed for firmly embedded broncholiths or peribronchial broncholithiasis, airway distortion, bronchiectasis, massive hemoptysis or fistula formation<sup>6,7</sup>. Broncho-lithotripsy is an alternative management for patients failing to remove broncholiths by surgery or bronchoscopy<sup>8,9</sup>.

General managements for hemoptysis, such as keeping airway patency, localization of bleeding, bleeding site down, coagulopathy correction, are the same in broncholithiasis related massive hemoptysis. Nonetheless, there are some different considerations in arterial embolization. Due to the fact that inflammation, hypoxia, and neoplasm promote bronchial arteries development<sup>10</sup>, 90% of massive hemoptysis originated from the bronchial arteries. So bronchial artery embolism is useful

management<sup>11</sup>. However, broncholithiasis does not induce such severe inflammation and may cause paucity of extravasation at angiography. Bleeding from the erosion of bronchololiths to adjacent pulmonary arteries may be the reason for massive hemoptysis secondary to broncholithiasis<sup>12</sup>. It might be the cause in our patient because of less mucosal inflammation at bronchoscopy and the adjacency of bronchololiths to the right pulmonary artery at CT (Figure 3A & 3B). The bronchial artery embolization, which was commonly used for malignancy related massive hemoptysis, may fail in this patient. Surgical resection may be considered for persistent bleeding resulting from broncholithiasis.

***Conclusion:***

Broncholithiasis may mimic malignancy at imaging and misdiagnosis may lead to different strategies for management of massive hemoptysis. Erosion of bronchololiths to adjacent pulmonary artery may be the reason for massive hemoptysis secondary to broncholithiasis, which may lead to failure of bronchial artery embolization. Clinician should keep in mind that broncholithiasis is a possible differential diagnosis for calcified mass presented with hemoptysis.

## **Reference**

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*Figures and figure legends*

Figure 1

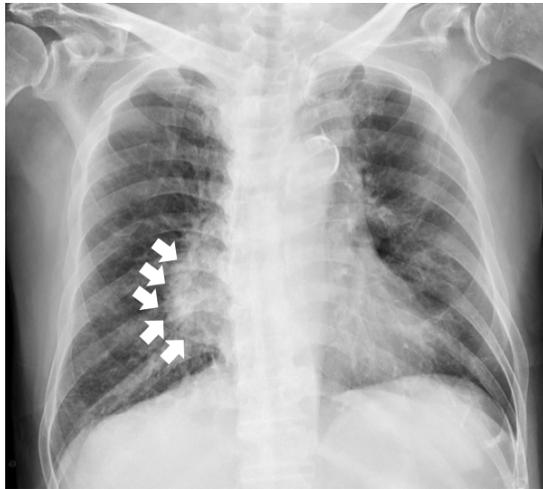


Figure 1

**Broncholithiasis as a mass on CXR in a 71-year-old man with fatal hemoptysis.**

A mass beside the heart border at CXR.

Figure 2

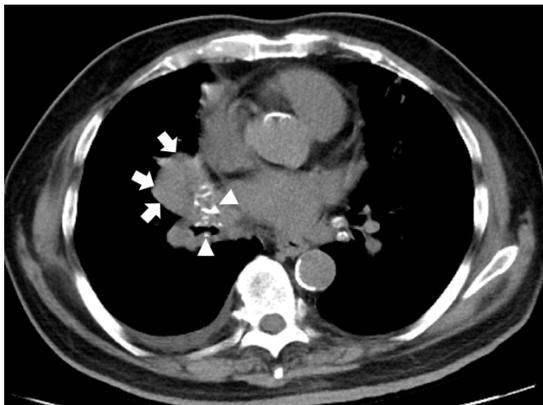


Figure 2

**Broncholithiasis as a mass with calcification on CT in a 71-year-old man with fatal hemoptysis.**

CT revealed a peribronchial mass with calcification.

Figure 3A

3B

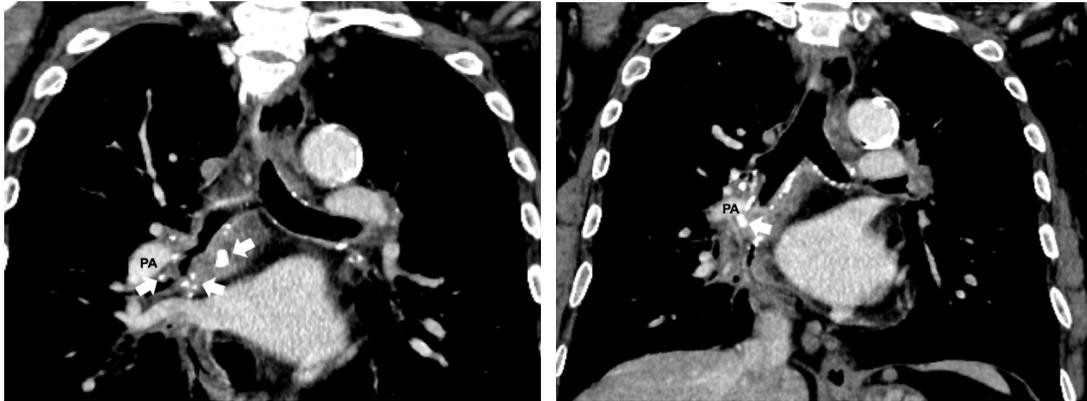


Figure 3

**Broncholiths protruded between right intermediate bronchus and right pulmonary artery in the patient with fatal hemoptysis.**

CT revealed broncholiths was around the right intermediate bronchus and protruded to right pulmonary artery, which may be the cause of life-threatening hemoptysis.