Recurrent Life Threatening Massive Hemoptysis as a Presentation of Pulmonary Actinomycosis

Kuo-Chin Kao, MD; Yao-Kuang Wu, MD; Chung-Chi Huang, MD; Meng-Jer Hsieh, MD; Ying-Huang Tsai, MD

Actinomycosis is a rare and slowly progressive infectious disease with mild symptoms such as low-grade fever, cough, chest pain and minor hemoptysis. We report an 81-year-old woman who was admitted to our intensive care unit with presentation of massive hemoptysis associated with hypotension, bradycardia and respiratory failure. The initial diagnosis was necrotizing pneumonia using chest X-ray and chest computed tomography. Another two episodes of life threatening massive hemoptysis with hemodynamic compromise occurred during treatment with antibiotics. Pulmonary angiography with embolization was attempted to stop the bleeding, however, it failed. Surgical intervention was performed due to uncontrolled recurrent life-threatening massive hemoptysis. Pulmonary actinomycosis was diagnosed during histopathology examination following unilateral pneumonectomy and bronchoplasty. The patient was successfully weaned from mechanical ventilator after surgery and was treated with intravenous Penicillin-G for 3 weeks after surgery. This patient was discharged without complications. Thus, recurrent life threatening massive hemoptysis should be considered as a presentation of pulmonary actinomycosis and surgical intervention may be necessary in selected cases. *(Chang Gung Med J 2006;29(4 Suppl):66-70)*

Key words: pulmonary actinomycosis, massive hemoptysis, pneumonectomy.

Actinomycosis is a chronic suppurative infectious disease, in which *Actinomyces* spp. infiltrates tissues associated with mucosal surfaces.[1] The disease usually presents in the cervicofacial and abdominopelvic forms and, less commonly, in the pulmonary form. Pulmonary involvement may in fact be misdiagnosed as a bronchopulmonary disease such as chronic infection or lung cancer, based on the chronic clinical course, poor response to antibiotic treatment, and radiographic presentation.[2]

In the majority of cases of clinical actinomycosis, the invasiveness of Actinomyces is greatly enhanced by a synergistic interaction with other bacteria, such as Staphylococcus, Streptococcus, and anaerobic bacteria.[3] Because of this polybacterial etiology, the correct diagnosis is often only made at the time of surgical intervention.[4,5] One of the hallmarks of pulmonary actinomycosis is the coughing up of blood from the respiratory tract. Hemoptysis is typically more frightening than life threatening. However, massive hemoptysis can cause a decrease in alveolar gas exchange and unstable hemodynamic conditions, and thus warrants prompt medical attention.

When confronted by massive hemoptysis, the major challenges facing clinicians are to establish the site or cause of the bleeding.[6] In order to provide more information for the clinician, we present an

From the Division of Pulmonary and Critical Care Medicine, Chang Gung Memorial Hospital, Taipei.
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Correspondence to: Dr. Ying-Huang Tsai, Division of Pulmonary and Critical Care Medicine, Chang Gung Memorial Hospital. 5, Fushing Street, Gueishan Shiang, Taoyuan, Taiwan 333, R.O.C. Tel.: 886-3-3281200 ext. 2281; Fax: 886-3-3287787; E-mail: chestmed@adm.cgmh.org.tw

unusual case of pulmonary actinomycosis with recurrent life threatening massive hemoptysis. The aetiological diagnosis was confirmed using pneumonectomy and histopathology examination results.

CASE REPORT

An 81-year-old woman had history of dry cough for 1 month, left-sided thoracic pain and dyspnea. She denied other medical disease such as autoimmune disease except for hypertension for 3 years and cerebral vascular accident with right hemiplegia for 2 years. She presented to the emergency room with two episodes of hemoptysis with fresh blood in amount of 20 to 30 ml in 1 day. After admission, physical examination revealed that her conjunctiva was pale and left breathing sound was wheezing. Her temperature was 36.2°C, heart rate was135 beats/min, respiratory rate was 22/min, and blood pressure was 138/78 mm Hg. The rest of the physical examination results were unremarkable including the chest wall was intact. Laboratory study results revealed a hemoglobin level of 6.5 g/dL, hematocrit of 20.5%, white blood cell count of 26200/μL, and a platelet count of 443000/μL. She was immediately transfused with 4 units of packed red blood cells for severe anemia. A coagulation profile was within normal limits.

On the admission day 1, she was intubated with a mechanical ventilator (MV) support because of an episode of life threatening massive hemoptysis with hypotension (87/32 mm Hg), bradycardia (heart rate 56 beats/min) and change of consciousness. A chest radiograph showed an extensive consolidation in the left lung (Fig. 1). The chest computed tomographic scan (Fig. 2) revealed that this consolidation was a left upper lobe atelectasis with air-bronchogram and cavitation due to necrotizing pneumonia.

With care in the ICU, the massive hemoptysis subsided and the hemodynamics stabilized. Through the endotracheal tube, bronchoscopic examination showed that there is no active bleeder or endobronchial mass from the lower trachea to the bilateral accessible bronchi. Some freely moveable blood clots in the anterior segment of the right upper lobe and much purulent secretion in the left lower lobe segmental bronchi were noted. No further procedure or examination (including biopsy) was performed. Microbiological examination of sputum failed to detect bacteria, mycobacterial tuberculosis or fungal infection. The patient was extubated 4 days after admission.

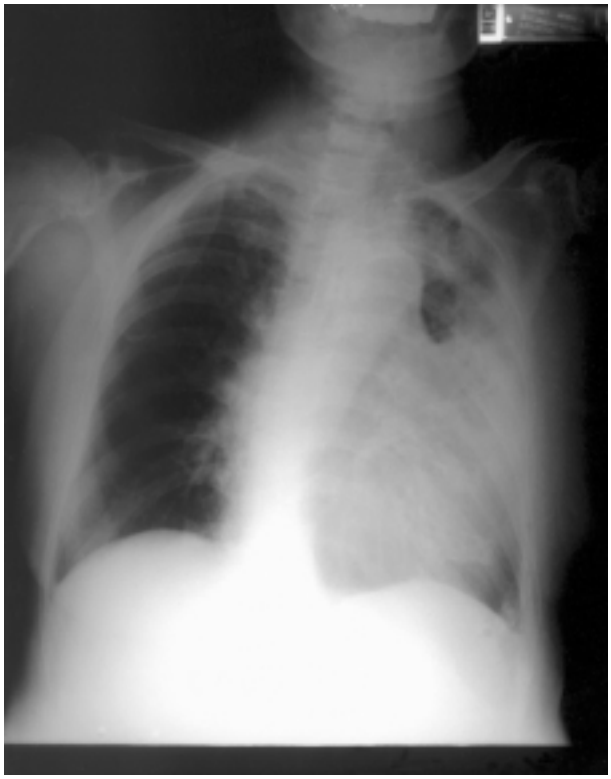

Fig. 1 Chest x-ray shows consolidation change in the left lung.

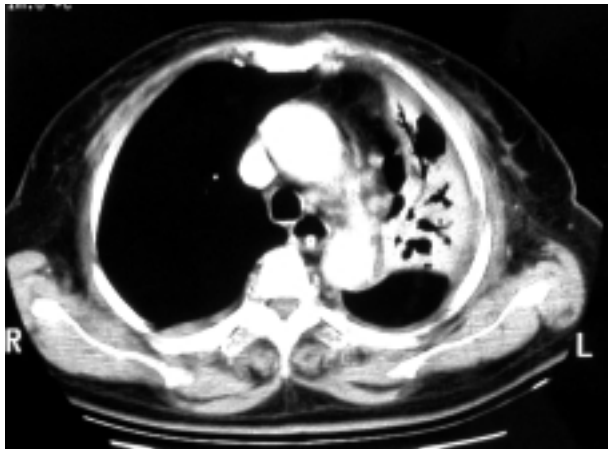

Fig. 2 Chest CT scan demonstrates left upper lobe atelectasis with air-bronchogram and cavitation due to necrotizing pneumonia.

Unfortunately, 3 days after extubation, the

patient had another episode of life-threatening massive hemoptysis with hypotension and she was intubated with MV support again. Radiologist was consulted for angiography with embolization but the bleeding stopped after intubation. Repeated bronchoscopic examination revealed no active bleeding in the visible bronchi. Quantitative microscopic cultures (including anaerobic cultures) of bronchoalveolar lavage yielded negative results. Results of fungal and mycobacterial smears and cultures were all negative. Cytological examination results of bronchoalveolar lavage from the left upper lobe was negative. Bronchial biopsy was not performed because there were no visible endobrochial masses. However, bacterial cultures of sputum were now positive for *Acinetobacter baumannii*. Despite intubation with MV support the patient experienced a third episode of life-threatening massive hemoptysis. A left total pneumonectomy was performed. Histopathology showed severe fibrosis and chronic inflammation of the lung in some segments. As well, "sulfur granules" and ulceration of the mucosal tissue were apparent in the airway (Fig. 3). After the third episode of hemoptysis, a final diagnosis of pulmonary actinomycosis was made.

Hemoptysis did not reoccur following surgery, and the patient was weaned from the mechanical ventilator smoothly. Penicillin-G was administered intravenously for the *Actinomyces* infection (12 million units daily) for 3 weeks. After discharge, oral antibiotics were administered for 4 weeks.

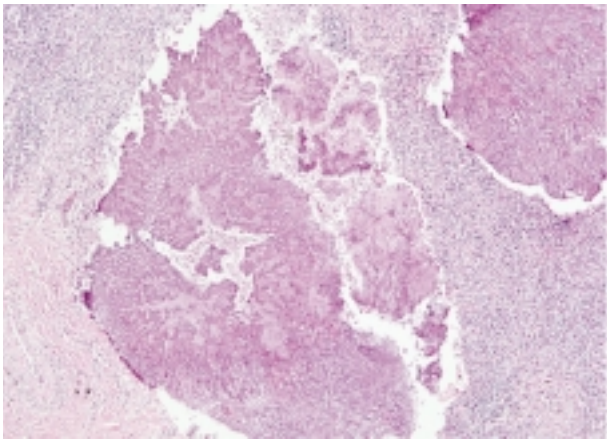

Fig. 3 Photomicrograph of lung tissue shows a large "sulfur granules" (arrow) in the small airway surrounded by abundant inflammatory cells. The bronchial mucosa shows marked erosion. (H & E stain, 40 X)

DISCUSSION

Actinomyces spp. is slow growing, filamentous, gram-positive, anaerobic bacteria that are typically part of the microbial population in the oropharynx and gastrointestinal tract. For example, the bacteria are found in 30-50% of normal saliva specimens.[3] *Actinomyces* spp. can cause the infection known as actinomycosis. Actinomycosis classically involves the mucosal tissue of the cervicofacial region (55% of cases), abdominopelvic region (20% of cases), thoracic region (15% of cases), and multiple organs (10% of cases).[7] Clinical symptoms frequently include fever, chills, hemoptysis, cough, pleuritic chest pain and anemia. In severe cases, it may present as empyema thoracis, empyema necessitatis, chronic draining sinus tract with typical "sulfur granules" contents, superior vena cava syndrome, or pericardial effusion.[9,10]

Pulmonary actinomycosis is thought to result from the entry of the bacteria into the thorax from the bronchial tree, either by inhalation of contaminated aerosol particles or by aspiration of contaminated matter from the upper digestive tract. In this cerebral vascular accident patient, the poor oral hygiene due to easy aspiration may explain the possible cause of pulmonary actinomycosis infection. The bacteria are always present synergistically with other opportunistic bacteria.[8] Actinomycosis is rare nowadays, because of better oral hygiene and the more extensive use of antibiotics. Nonetheless, pulmonary actinomycosis can prelude a wide variety of pulmonary diseases including unresolved or poorly responding pneumonia, infarction, nocardiosis, tuberculosis, bronchogenic carcinoma, cryptococcosis, and histoplasmosis.[11,12]

Classically, thoracic actinomycosis is parenchymal infiltrate with involvement of the chest wall with ribs eroded.[13] Typical chest x-ray findings are not specific. Chest computed tomographic scanning usually reveals a soft tissue mass with varying degrees of infiltration, abscess formation and pleural thickening adjacent to the airspace consolidation.[14,15] The demonstration of the bacterial origin of pulmonary actinomyces is complicated by the failure to conduct a bacteriological examination of the sputum in many cases, or because the culture was performed only in the presence of oxygen. The latter conditions do not

allow for the growth of *Actinomyces* spp., which tolerate only low levels of oxygen. Therefore, accurate diagnosis of primary pulmonary actinomycosis is rarely made at the time of admission and is frequently made using histopathology rather than by microbiological methods.[2]

The actinomyces pathogens are not especially virulent, and so the symptoms of low-grade fever, cough, chest pain and hemoptysis are not typically life threatening.[8]

Endobronchial actinomycosis is another form of actinomycosis which presents with endobronchial masses and hemoptysis.[14,15] Hemoptysis occurred in the pulmonary region. The overall mortality rate attributed to massive hemoptysis is largely influenced by malignant etiologies and by the bleeding rate. Conservative medical therapy may suffice in certain conditions and other interventions (such as endobronchial tamponade, bronchial artery embolization or surgery in eligible candidates) should be undertaken if the bleeding fails to stop after conservative measures. Actinomycosis is more frequently seen than endobronchial actinomycosis (77% in pulmonary actinomycosis).[7] Therefore, the risk of life-threatening massive hemoptysis is higher in patients with pulmonary actinomycosis than those with endobronchial actinomycosis. Hemoptysis is much more often attributable to chronic inflammatory lung disease and bronchogenic carcinoma in the United States. Tuberculosis continues to be the leading cause of hemoptysis worldwide.[16] The most common etiologies of massive hemoptysis are neoplasm, bronchiectasis, infections, vascular disease, and vasculitis.[6]

The surgical indication of pulmonary actinomycosis is limited to diagnostic purposes.[2] Surgical management should be considered when response to penicillin therapy is poor and hemoptysis is not resolvable.[7] In this presenting case, surgical intervention was performed due to uncontrolled recurrent life-threatening massive hemoptysis. The cause of delayed surgical intervention of life-threatening massive hemoptysis was the hemoptysis had resolved quickly in the previous two episodes. Histopathological examination conducted during the course of the episodes of hemoptysis revealed actinomycosis with sulfa granules, which is characteristic of an *Actinomyces* infection. When the bacterium was finally isolated from the sputum, the appropriate antibiotic treatment began. This strategy proved to be successful.

In conclusion, the case study presented here is consistent with the suggestion that recurrent life-threatening massive hemoptysis may be a rare presentation of pulmonary actinomycosis. Surgical intervention and antibiotic treatment with penicillin for 6 months may represent a useful treatment of this condition.

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肺部放線菌病以反覆危及生命的大咳血來表現

高國晉 吳燿光 黃崇旂 謝孟哲 蔡熒煌

放線菌病是一種少見且緩慢進行而伴有輕微症狀，例如；輕微發燒、咳嗽、胸痛和少量咳血的感染症。我們報告一例八十一歲女性病患因大量咳血合併有低血壓、心跳過慢以及呼吸衰竭而住進加護病房；經由胸部 X 光及電腦斷層掃描檢查，初始診斷爲壞死性肺炎，然而在以抗生素治療的當中又發生兩次合併有血液動力學變化的大量咳血，由於反覆危及生命的大量咳血，因此施以肺動脈血管攝影栓塞法試圖止血，但失敗，因此接受手術單側肺切除來治療，最後經由組織病理學的檢查，確定診斷爲肺部放線菌病。術後再經由三周的靜脈注射盤尼西林抗生素治療，病患成功脫離呼吸器，並且無其他併發症的出院。因此，反覆危及生命的大咳血可以是肺部放線菌病的表徵，外科手術治療在某些病患是須要的。(長庚醫誌 2006;29(4 Suppl):66-70)

關鍵字：肺部放線菌病，大量咳血，肺切除。

長庚紀念醫院 台北院區 胸腔科
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索取抽印本處：蔡熒煌醫師，長庚紀念醫院 胸腔科。桃園縣333龜山鄉復興街5號。Tel.: (03)3281200轉2281；Fax: (03)3287787; E-mail: chestmed@adm.cgmh.org.tw