Int J Clin Exp Med 2016;9(11):22538-22542
www.ijcem.com /ISSN:1940-5901/IJCEM0035626

Case Report

Behcet's disease with a pulmonary mass: tumor or infection?

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Received July 12, 2016; Accepted September 1, 2016; Epub November 15, 2016; Published November 30, 2016

Abstract: Pulmonary actinomycosis with haematogenous dissemination is an extremely rare situation occurred after autoimmune disease. Here we report an interesting case of Behcet's Disease (BD) in a 41-year-old man presenting with swelling and painful right wrist joint. At first, with the improvement of the imaging tests, it suggested lung mass indicating pulmonary cancer with bone metastases. Completed microbiological and pathological examinations showed it was pulmonary actinomycosis and hematogenous dissemination to the hand, similar to the bone metastasis of lung cancer finally.

Keywords: Behcet's disease, pulmonary actinomycosis, hematogenous dissemination

Introduction

Actinomycosis is a chronic suppurative or granulomatous infection, which most commonly occurs in 3 body regions: cervicofacial (65%), abdominopelvic (20%), and pulmonothoracic (15%) [1]. Due to the use of hormone, cytotoxic drugs and immunosuppressive agents, the reports of lung action mycosis increases recent years [2]. However, Actinomycosis with haematogenous dissemination is a rare disease. We present a rare case of pulmonary actinomycosis and hematogenous dissemination confirmed with a biopsy.

Case report

A 41-year-old man visited our hospital complaining of the right wrist joint with swelling and pain for 2 days. The patient had suffered Behcet's Disease (BD) for over 10 years, and had been taking oral hormone and immunosuppressive agents as required. He had no oral and genital sores, fever, cough, sputum and chest pain recently. On physical examination, vital signs were normal. There was a low and unclear respiratory sound on the right upper lung, without dry and wet rale. The right wrist joint was swelling and tenderness, along with the increased skin temperature.

Laboratory data were notable for a leukocyte 2.17×10^9/L (Reference range, 3.5-9.0×10^9/L), neutrophils rate 65.54% (Reference range, 50-70%), ultrasensitive C-reactive protein 98.24 mg/L (Reference value, 0-10 mg/L), erythrocyte sedimentation rate 8.84 mg/L (reference value, 0-5 mg/L), procalcitonin 0.155 ng/ml (reference value 0-0.046 ng/ml), respectively. IgG, IgA, IgM, IgE, C3 and C4 were all within normal value. There were no abnormalities in other serological tests. Due to increased inflammatory marker, Behcet's disease involved arthritis could not be ruled out completely.

However, a chest CT scan on the right upper lung revealed a dense lump, indicating the possibility of a lung tumor. For a definitive diagnosis, we conducted PET/CT scan for the patient and the results showed a 7.0×8.5 cm mass in the upper lobe of the right lung with soft tissue density and speculated margins, which was intense hypermetabolism as a tumor (**Figure 1**). The bone scan found the uptake of radiotracer was high signal at the right hand wrist (**Figure 2**). Combined with the above imaging tests, we considered the possibility of lung cancer with bone metastases. Next, A CT-guided lung puncture biopsy was implemented to confirm the nature of the mass, which showed purulent inflammation and actinomycetes (**Figure 3**). A

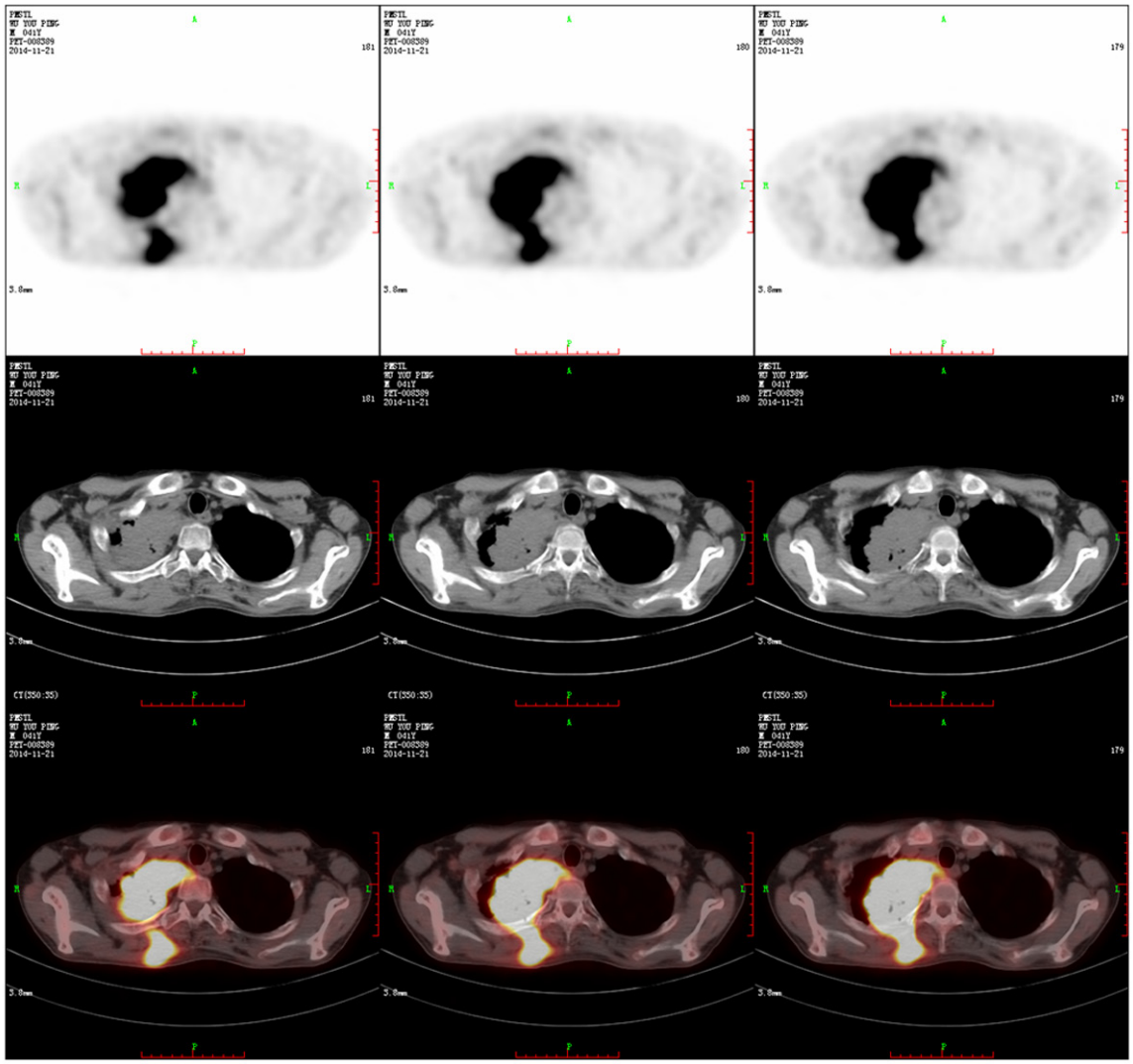

Figure 1. 41-year-old man with pulmonary actinomycosis imitating lung cancer with PET/CT scan. The PET/CT showed a 7.0×8.5 cm mass in the upper lobe of the right lung, accompanied by increasing glucose metabolism.

few days later, there was a fluctuation on the patient's right wrist. A little yellowish pus was collected by puncture the fluctuation after sterilization. Dental caries actinomycosi was cultured from pus of biopsy spot. Taking the clinical and histological presentation into consideration, the final diagnosis was made: pulmonary actinomycosis and hematogenous spread to right wrist.

With a treatment of intravenous antibiotics (Piperacillin sodium and Sulbactam combined with levofloxacin hydrochloride) for 15 days, the CT test showed the size of mass was decreased (**Figure 4**). Meanwhile the swelling of right wrist joint was reduced gradually after puncture drainage. Cephalosporines were prescribed to be taken for 3 more months. There was effective improvement on the patient's physical condition.

Discussion

The present case is characterized by a swelling and painful joint as its first manifestation, but without recurrent oral and genital sores, fever, cough, sputum and chest pain. The diagnostic process is complicated. Combined with the history of BD and increased inflammatory markers, relapse of BD associated arthritis was considered when admitted to hospital. With the improvement of the imaging tests, it suggested that lung cancer with bone metastases. Finally, it diagnosed as actinomycosis by microbiological and pathological examinations.

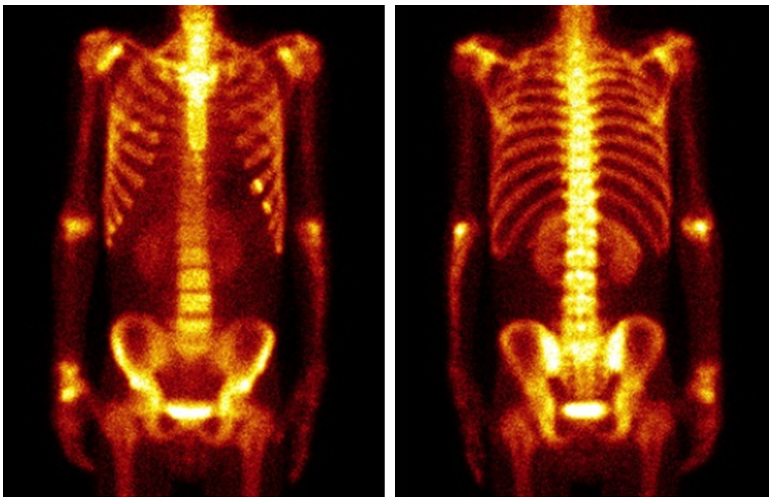

Figure 2. The bone scan found the uptake of radiotracer was high signal at the right hand wrist.

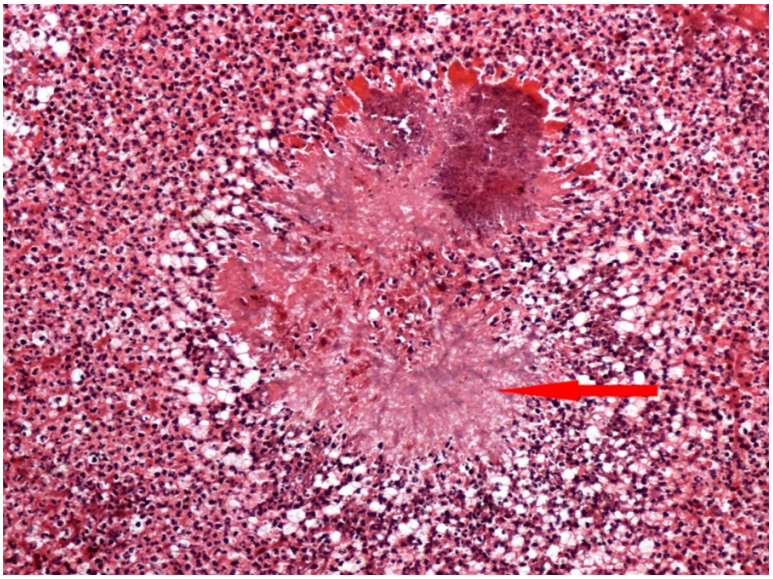

Figure 3. Bacterial colony in the abscess background showing actinomycosis filamentous organisms. (H&E, ×200).

Actinomycosis is a chronic suppurative or granulomatous infection caused by Actinobacteria, and the incidence is 1/300,000 [3]. The common clinical symptoms include fever, chest pain, low-grade temperature, hemoptysis, pyothorax. Cough, expectoration and haemoptysis also occur frequently [4]. Recently, many reports reveal that actinomycosis is an opportunistic infection [5, 6]. The people who suffer from human immunodeficiency virus (HIV), tuberculosis (TB) and Connective Tissue Disease (CTD) using high dose of hormone, cytotoxic drugs or immunosuppressive agents are more easily infected with actinomycetes than normal [2, 7]. When the secretion containing actinobacteria on the throat is breathed into the lower respiratory tract, it can cause primary lung actinomycosis. Owing to the long-term use of oral immune inhibitors and hormones, the man is at high risk of opportunistic infection in our case. The patient has not any typical symptoms of infection after admission, so the diagnosis is delayed. This is inconsistent with the report that 94 cases of pulmonary actinomycosis have common symptoms of cough, expectoration and hemoptysis [4]. We must pay attention to the special infection occurred when the primary disease is autoimmune disease.

The actinomycosis is developed mainly through direct infiltration spreading. Haematogenous dissemination of the disease is rare. Review of the literature, few case reports about lung actinomycetes to the distant spread are found. Lin Qiu reports a case of 41-year-old man of pulmonary actinomycosis with multiple abscesses [8]. In our case, in addition to the direct infiltration of spread, the man is infected by blood line which is confirmed by culturing pus of wrist. Although he diagnosed as pulmonary actinomycosis and distant metastasis to wrist, he has no obvious evident of poisoning symptom. It may be related to the bacterial strain of low toxicity and the use of hormones, which cover the disease.

Meanwhile, clinical manifestations and imageological examination in pulmonary actinomycosis are nonspecific, which is often raising suspicion of a malignant cancer [9]. Therefore,

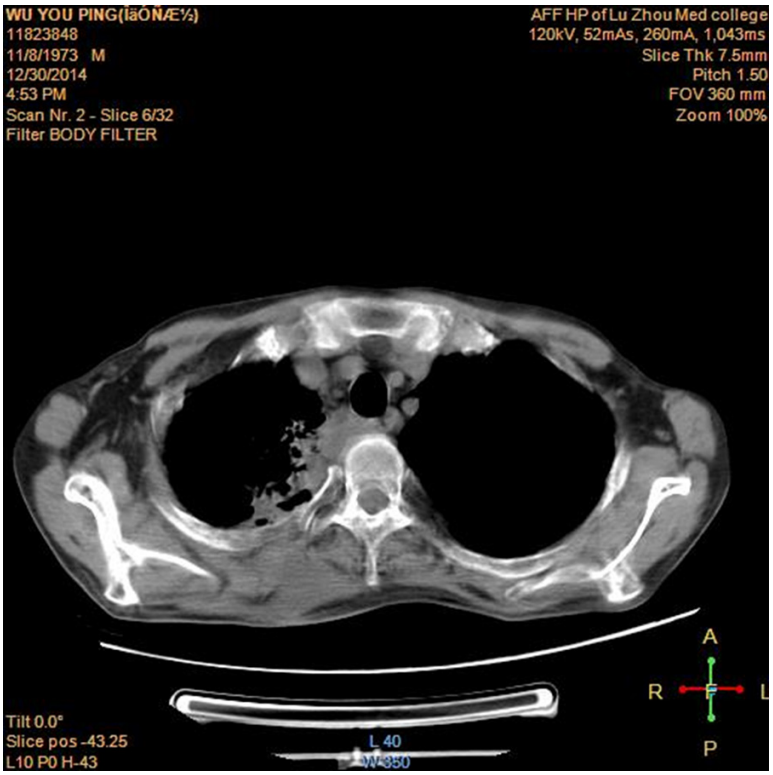

Figure 4. The chest CT showed the size of mass was decreased evidently.

as with our case, establishing an accurate diagnosis may be difficult and significantly prolonged [10]. The general PET/CT image of actinomycosis is intense hypermetabolism which is hard to differentiate from malignancy [11]. PET/CT examination can show a densed roundish and sheet like image on the right upper lobe section, accompanied by increasing glucose metabolism, which is consistent with the report of Lee finding high absorbing of 18F-fluorodeoxyglucose (18F-FDG) in the lung actinomycetes lesions [12]. Clinical utilization of PET/CT is an examination to distinguish between benign and malignant tumors. It can provide morphology and metabolic activity of lesions, so as to offer the help for diagnosis. Due to lesions of metabolic activity, 18F-FDG is a large amount of intake, which is proportional to the number of malignant tumor cells and proliferation activity [13]. However, pulmonary inflammatory lesions can be a high intake of 18F-FDG, which is not mean to the metabolic activity but the existence of white blood cells and the activity. The principle of this assumption is the activation of macrophages and neutrophils require large amounts of glucose as a power source to complete the chemotaxis and phagocytosis in inflammatory tissue [14]. Therefore, there is no certain value to identify malignant tumor and actinomycosis by the PET/CT. Although PET/CT indicates the lamp is likely to be malignant tumor in the present case, ESR, CRP and PCT increase significantly. In conclusion, when neoplasm is highly suspect using PET-CT scan, we should be careful differentiate with inflammatory disease, especially inflammation index increased. If the diagnosis is elusive, we can do a tissue biopsy to make a definitive diagnosis.

Disclosure of conflict of interest

None.

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