Pulmonary Actinomycosis Imitating Malignancy in an Epileptic Patient

Epileptik Bir Hastada Maligniteyi Taklit Eden Pulmoner Aktinomikoz

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ABSTRACT

The diagnosis of pulmonary actinomycosis is still a difficult problem in medical practice. A 38 year old female who had been using antiepileptic drugs for 27 years was admitted to our hospital with complaints of cough and yellowish sputum of six months duration. Physical examination of the patient was unremarkable. Chest radiography showed a lesion involving the pleura of the right hemithorax. Thorax CT revealed a hypodense mass. Fiberoptic bronchoscopic examination and transbronchial biopsy were not diagnostic. The patient was diagnosed as pulmonary actinomycosis after diagnostic thoracotomy. In the present report, we concluded that pulmonary actinomycosis must be kept in mind in the differential diagnosis of an epileptic patient with the radiological findings of malignancy.

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Key words: Actinomycosis, aspiration, epilepsy

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ÖZET

Pulmoner aktinomikoz tanısı klinik pratikte hala önemli bir sorundur. Yirmi yedi yıldır antiepileptik tedavi kullanan 38 yaşındaki bayan hasta 6 aydır süren öksürük ve sarı balgam ekspektorasyonu yakınmaları ile kliniğimize başvurdu. Fizik muayenede özellik yoktu. Akciğer grafisinde lezyonun plevra ve sağ hemitoraks ile ilişkili olduğu izlendi. Toraks BT'de hipodens kitle saptandı. Fiberoptik bronkoskopik inceleme ve transbronşial biyopsi tanısal değildi. Tanısal torakotomi ile hastaya pulmoner aktinomikoz tanısı kondu. Bu yazıda malignite şüphesi olan epileptik hastaların ayırıcı tanısında pulmoner aktinomikozun da bulunması gerektiği vurgulanmıştır.

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Anahtar sözcükler: Aktinomikoz, aspirasyon, epilepsi

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INTRODUCTION

Actinomycosis is a rarely seen chronic suppurative infection which is caused by Gram positive, non-spore forming, predominantly anaerobic precaryotic bacteria called *Actinomyces*. The classical form of the disease is the cervicofacial type in which a middle aged patient presents with a large mass on the jaw. However, the disease can affect every system in the human body. Besides the cervicofacial type (50%); pulmonary (15%), abdomino-pelvic (20%), central nervous system (2%), cutaneous, cardiac and genitourinary involvements were also reported in literature. Patients who are alcoholic and have poor oral hygiene, facial trauma, diabetes mellitus, underlying respiratory diseases such as emphysema, chronic bronchitis and bronchiectasis are susceptible to the disease [1]. Besides all these risk factors, our case suggests that epilepsy may be taken as a risk factor for pulmonary actinomycosis [2]. Diagnosis of actinomycosis is frequently difficult because it often mimics tuberculosis, lung abscess or lung cancer clinically and radiologically [3-4]. In the present study, we discuss a pulmonary actinomycosis case in the light of the literature.

CASE

A 38 year old female patient was admitted to our hospital with the complaints of cough and yellowish and sometimes bloody sputum for the previous six months. It was learned that the patient had been using Carbamazepin and Levatiresetam for epilepsy since she was 11 years old. Anti-tuberculosis therapy had been given in another center with the diagnosis of smear negative pulmonary tuberculosis. Anti-tuberculosis therapy was terminated on the fifteenth day of the treatment because of hepatotoxicity. When the patient was admitted to our center, she had not had anti-tuberculosis therapy for 2 months.

The physical examination of the patient was unremarkable. Laboratory studies revealed a high sedimentation rate of 56 mm/h. The results of CBC, blood chemistry and urine analysis were all within normal ranges. Chest radiography showed a lesion involving the pleura

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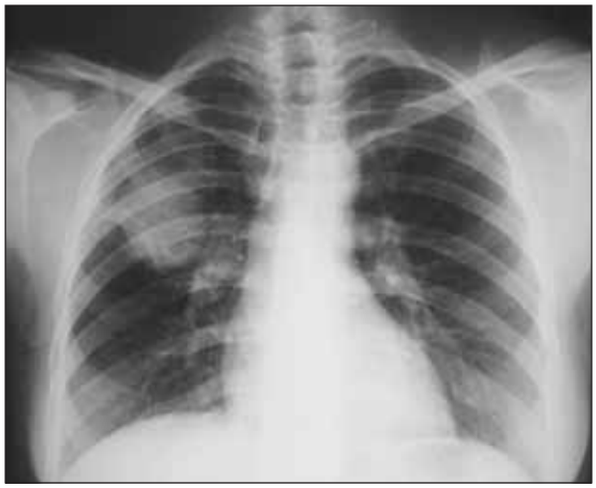

Figure 1. Chest radiography showed a mass in the right hemithorax

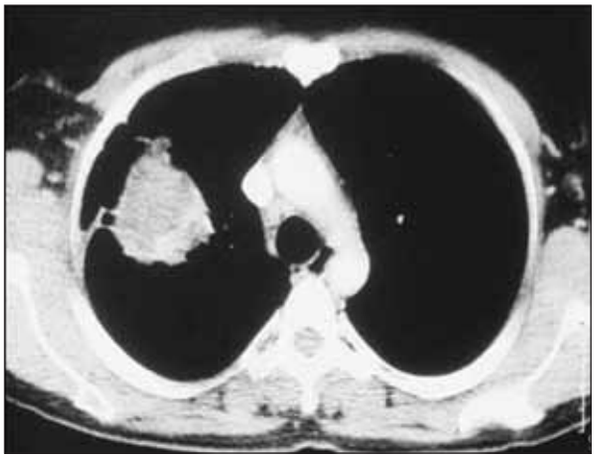

Figure 2. Thorax CT showed a mass in the upper lobe of the right lung

on the middle zone of the right hemithorax (Figure 1). In her thorax CT, there was a cystic- hypodense mass of 4.5-5.0 cm in size with irregular borders in the upper lobe of the right lung (Figure 2). Cranial CT and abdominal USG were normal. At the fiberoptic bronchoscopic (FOB) examination there was no endobronchial lession and transbronchial biopsy under scopy was performed from the apical segment of the right upper lobe. Pathologic examination of the transbronchial biopsy revealed inflamed bronchial mucosa and FOB lavage was reported as blood components. As the transbronchial biopsy taken via FOB was not diagnostic, the patient underwent diagnostic thoracotomy. After right thoracotomy, a frozen section biopsy examination revealed parenchymal tissue containing hemorrhagic focuses and infiltrated fibrosis with dense inflammatory areas The samples of the content of the cyst showed colonization of actinomyces. In the histopathologic examination basophilic sulphur granules with radial eosinophilic halo were seen in a necrotisng inflammatory area (H.E.; X100) (Figure 3). Thereby, the histopathologic diagnosis of pulmonary actinomycosis was achieved after diagnostic thoracotomy. The patient was given 24 million units of penicillin G treatment for three months and then it

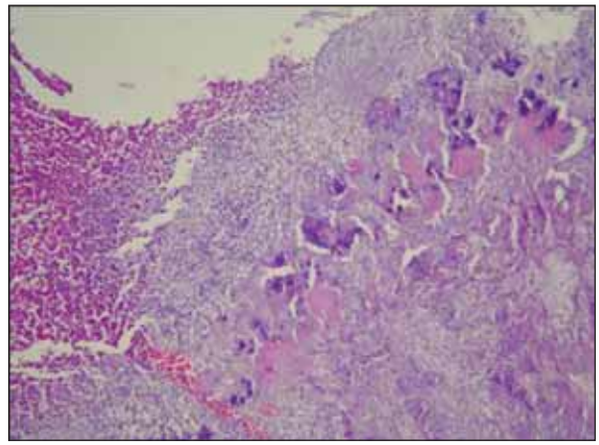

Figure 3. Basophilic sulphur granules with radial eozinophilic halo are seen in a necrotising inflammatory area (H.E.; X100)

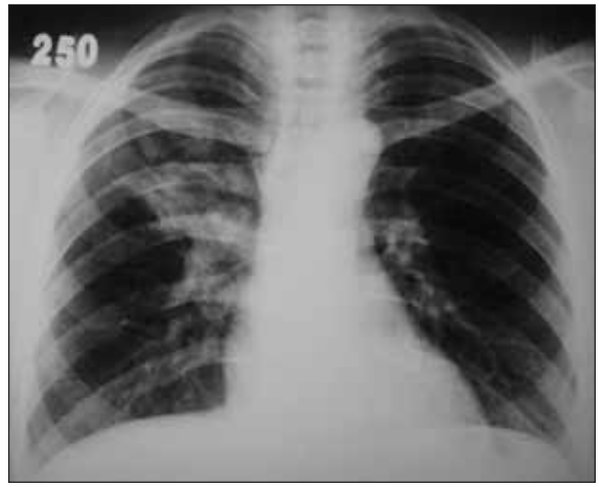

Figure 4. Chest radiography showed radiological regression in the third month of treatment

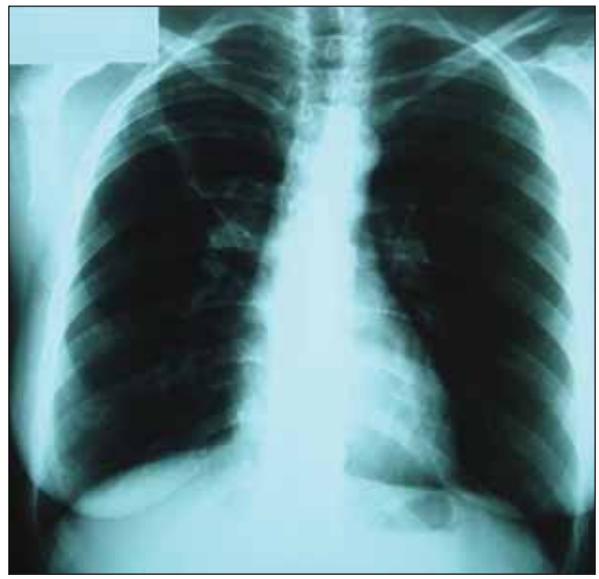

Figure 5. Chest radiography showed radiological regression at the end of treatment.

was planned to complete treatment for six months with oral penicillin V.

After the end of the third month a clear radiological regression was achieved (Figure 4). At the end of the treatment after nine months, the disease had regressed completely (Figure 5).

DISCUSSION

Actinomycosis is a rarely seen chronic infectious disease which is caused by bacteria called *Actinomyces*. The disease has two peaks at the ages of 11-20 and more commonly in the 4th-5th decades. Males are affected 2-4 times more than women [1]. However, our patient was a woman in her late thirties.

The clinical suspicion of actinomycosis can arise from radiological findings and the history of the patient in combination. If the patient is an alcoholic or has poor oral hygiene, facial trauma, dental disease or underlying respiratory disorders such as emphysema, chronic bronchitis and bronchiectasis, then clinicians must be alert for signs of pulmonary actinomycosis. These risk factors support the aspiration based pathogenesis of the disease. Our patient had none of these risk factors but she had been epileptic for 27 years and was using Carbamazepin and Levatiresetam. Besides all these risk factors, our case showed that epilepsy must also be regarded as a risk factor for Pulmonary Actinomycosis. It was thought retrospectively that the aspiration of oropharyngeal and gastrointestinal flora of which actinomyces spp. are native members, could occur during her epileptic seizures. Although there are some new findings suggesting that aspiration pneumonia is not a common complication of seizures in otherwise healthy adults [2, 3], the classical textbooks still associate aspiration pneumonia in adults with generalized tonic-clonic seizures [4].

Characterized radiological findings can be helpful for diagnosis as well. Plain chest radiographs are nonspecific and can resemble lung cancers, or benign infections. In a study by Cheon JE at al, all the lesions had been found to be unilateral, with an average diameter of 6.5 cm. Patchy air space consolidation with central areas of low attenuation (75%) and adjacent pleural thickening (73%) at chest CT scans have been shown to be suggestive of thoracic actinomycosis [5]. Central areas of low attenuation had been correlated histopathologically as necrotic material containing enlarged bronchi or microabscesses with sulphur granules which are hallmarks of Actinomycosis. In our patient's CT scan, the lesion was unilateral and centrally hypodense with a diameter of 5 cm and adjacent pleural thickening. All these findings are in accord with the above-mentioned article.

Since Actinomyces are part of the normal respiratory flora, a positive sputum culture is of little clinical significance. If the diagnosis cannot be made by simpler ways, transthoracic needle aspiration, transbronchial biopsy or open lung biopsy should be performed [6]. Diagnosis of actinomycosis is frequently difficult because it often mimics tuberculosis, lung abscess or lung cancer clinically and radiologically. Therefore, surgery then provides the best method to achieve diagnosis. Beside all these clues for thoracic actinomycosis, the fact that the disease can be misdiagnosed as lung cancer most commonly and, furthermore, can coexist with lung cancer makes surgical diagnosis inevitable. To give a few examples from the literature, in Tastepe's series of 7 patients and Rizzi's series of 13 patients all cases were diagnosed via surgery except one [7, 8]. There are only a few patients reported to be diagnosed after bronchial biopsy or percutaneous fine-needle aspiration [1]. Our case underwent diagnostic thoracotomy after a non-diagnostic transbronchial biopsy and no complication was seen. Additionally, surgical intervention may be necessary for treatment as well when frequent hemoptysis has no resolution, or complications such as well defined abscesses, empyemas, discharging fistulas and sinuses are seen and lung neoplasm can not be ruled out, as in our patient [9].

After diagnosis, the disease requires prolonged antibiotic therapy because of the avascular, scarred, deep-seated nature of the infection. The recommended therapy for this infection is an intravenous course of appropriate antibiotics followed by oral treatment on an outpatient basis in conjunction with surgical intervention. ß lactam antibiotics are the drugs of choice. Generally 18-24 million units penicillin G for 2-4 weeks are applied initially and this is followed by oral penicillin or amoxycillin for 3-12 months in addition. In penicillin-allergic patients, tetracyclines can be an alternative. In pregnant patients, erythromycin is safe [1]. We administrated 3 months of intravenous penicillin (24 million units per day) and an additional 6 months of oral penicillin (2000 mgr per day) to our patient. Radiological regression was achieved in the third month of the therapy.

As the diagnosis of thoracic actinomycosis requires a combination of several factors, it is quite difficult to diagnose. However, it is curable when recognized early and the appropriate treatment is given. Our case underlines that epilepsy is a risk factor for thoracic actinomycosis. We recommend that when epilepsy coexists with the radiological findings mentioned above, thoracic actinomycosis should be kept in mind in the differential diagnosis. To avoid unwarranted surgery, diagnostic methods for thoracic actinomycosis merit further investigations.

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