



Unusual aetiology of lymphocyte-predominant exudative pleural effusion: primary mediastinal actinomycosis

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Keywords

¹⁸F-fluorodeoxyglucose uptake, mediastinal actinomycosis, pleural effusion.

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Abstract

Actinomycosis is a rare infectious bacterial disease typically involving cervicofacial, abdominopelvic, or thoracic regions, caused by *Actinomyces* species. However, involvement of the mediastinum as the primary site of origin is extremely rare. An elderly patient complained of left-sided chest pain. The chest radiograph revealed pleural effusion, which revealed lymphocyte-predominant exudates. Chest computed tomography showed a soft tissue mass in the left anterior mediastinum. Positron emission tomography revealed an increased uptake of ¹⁸F-fluorodeoxyglucose in this lesion. To exclude malignancy and make a confirmative diagnosis, a mediastinal biopsy was performed through video-assisted thoracic surgery. Finally, mediastinal actinomycosis was diagnosed. We report herein a case of mediastinal actinomycosis mimicking lung malignancy presented with recurrent lymphocyte-predominant pleural fluid exudate. In patients with a recurrent or unexplained exudative pleural effusion, it may be worthwhile to consider a hidden foci of actinomycosis.

Introduction

Actinomycosis is a rare, chronic, and suppurative granulomatous infection caused by facultative anaerobic Gram-positive bacteria of the genus *Actinomyces*. These pathogens typically cause three classical forms of actinomycosis (cervicofacial, thoracic, and abdominal), but can also cause infections of the female genitalia, eyes, and tissues adjacent to dental implantation elements and tooth extraction wounds [1]. Thoracic actinomycosis accounts for approximately 15% of all actinomycosis cases. Mediastinal involvement is extremely rare, except when related to the use of medical devices, such as oesophageal stents.

Case Report

A 67-year-old man with a history of hypertension was first admitted to our hospital with complaints of chest pain on the left side exacerbated by deep breathing. He reported that his symptoms had begun three weeks ago. He had quit

smoking 30 years ago and had a 15 pack-year history of smoking. A chest radiograph showed a small amount of left-sided pleural effusion (Fig. 1A). Therefore, thoracentesis and pleural fluid analysis were performed, and lymphocyte-predominant exudates were established (pH 7.5, white blood cells of 2800/ μ L, lymphocyte 93%, eosinophil 4%, monocyte 3%, glucose 126 mg/dL, protein 5.0 g/dL, and lactate dehydrogenase 291 IU/L). The level of pleural fluid adenosine deaminase was 17 IU/L and there were no malignant cells present. A soft tissue mass of about 1 \times 3.5 cm in the left anterior mediastinum was revealed by chest computed tomography (CT) (Fig. 1D). Surgical biopsy of the mediastinal lesion was recommended; however, the patient declined and was discharged. Four months after the initial visit, the patient returned and the left pleural effusion was moderately decreased (Fig. 1B). However, after another four months, the amount of left-sided pleural effusion had substantially increased, while the mass in the mediastinum had slightly decreased (Fig. 1E). Patient was transferred to

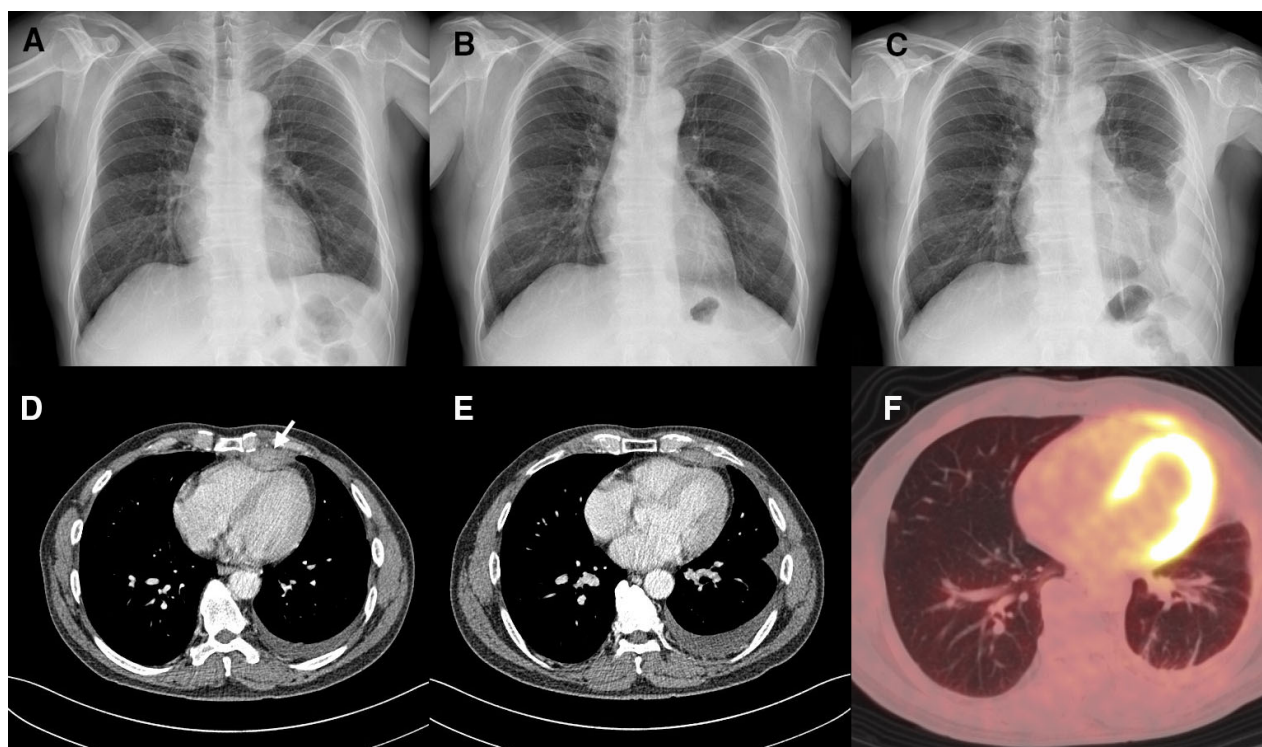


Figure 1. (A) An initial chest radiograph showing a small amount of left-sided pleural effusion. (B) A moderate decrease in the amount of left pleural effusion after four months. (C) A significant increase in the amount of pleural effusion after 14 months. (D) The initial CT image revealing a left anterior mediastinal mass (arrow) sized 1.0×3.5 cm and a left pleural effusion. (E) A decrease in mass and an increase in left pleural effusion after eight months. (F) Development of multifocal subpleural consolidations and atelectasis in the lingular segment and left lower lobe after 14 months and ^{18}F -fluorodeoxyglucose (FDG) uptake (maximum standardized uptake value (SUVmax): 6.3) was increased in the mass-like lesion at the left anterior mediastinum.

another hospital by the request of his caregiver and no further evaluation was conducted.

Fourteen months after the first visit, the patient was admitted back at our hospital complaining of left-sided chest pain exacerbated by deep inspiration. Pleural effusion had significantly increased (Fig. 1C) and a pigtail catheter was inserted into the left pleural space. A follow-up CT scan revealed newly developed multifocal subpleural consolidations and atelectasis of the lingular segment and left lower lobe. However, the extent of soft tissue density in the mediastinum did not show a significant change. Pleural fluid analysis still showed lymphocyte-predominant exudates (white blood cells of $1100/\mu\text{L}$, lymphocyte 82%, and adenosine deaminase 20 IU/L) and no malignant cells were visible in the cytological examination. Under the suspicion of malignancy, a positron emission tomography (PET) CT scan was performed. Intense ^{18}F -fluorodeoxyglucose (FDG) uptake was observed in the left anterior mediastinum, suggesting a malignant or metastatic lesion (maximum standardized uptake value (SUVmax) = 6.3). However, increased FDG uptake of pleural, atelectatic, and consolidative lung

lesions was unremarkable (Fig. 1F). Biopsies for the mediastinal mass and pleura were performed with a video-assisted thoracoscopic surgery. The specimen from mediastinal mass showed finely branching filamentous materials, which were positive for the periodic acid–Schiff staining method (Fig. 2). Pleural tissue showed thickening of the loose connective tissue layer and non-specific inflammatory cells. Therefore, the patient was diagnosed with mediastinal actinomycosis and secondary lymphocyte-predominant exudative pleural effusion caused by *Actinomyces* infection.

The patient was treated with intravenous amoxicillin/sulbactam for seven days and discharged with a prescription of oral amoxicillin/clavulanic acid. After completing three months of treatment, he did not present chest pain or demonstrable lesion of the mediastinum on the follow-up CT scan.

Discussion

Thoracic actinomycosis is believed to be caused by aspiration of the oropharyngeal contents, which leads to bronchopulmonary invasion of the organism. This may

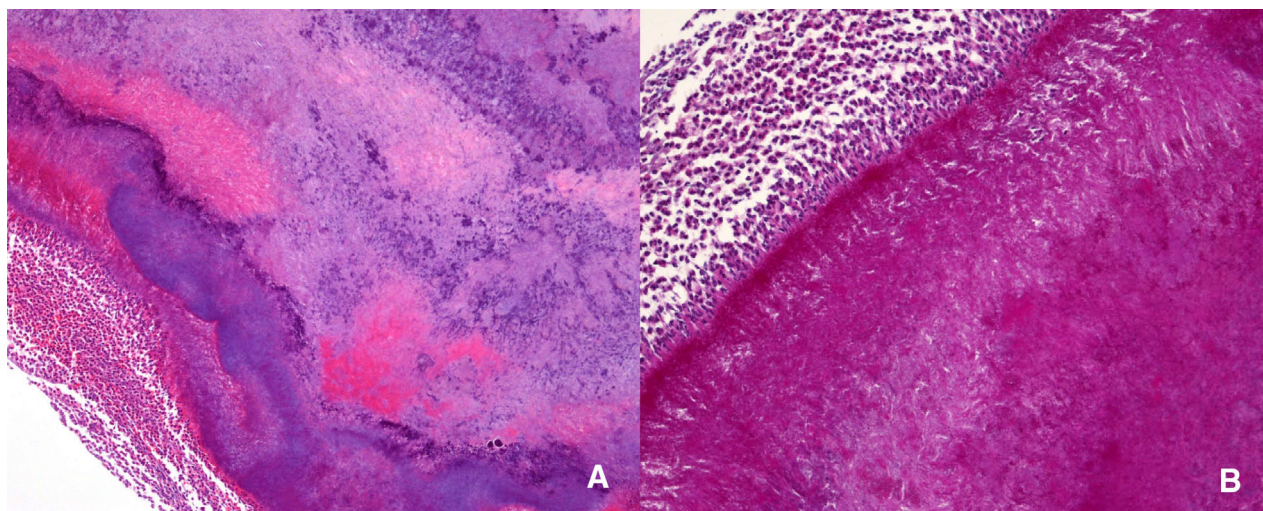


Figure 2. Histopathology showing (A) sulphur granules of *Actinomyces* (haematoxylin and eosin (H&E), 100x) and (B) Periodic acid-Schiff (PAS) staining positive for finely branching inner filamentous materials with circumjacent neutrophilic reactions (PAS, 200x).

subsequently spread to the chest wall and/or mediastinum [2]. However, no causative factor was observed in this patient. The presence of pleural effusion is also uncommon in patients with pulmonary actinomycosis [3]. There have been a few reports describing the characteristics of pleural or pericardial fluid caused by actinomycosis, which differ in the cellular distributions. For example, Coodley and Yoshinaka reported a case of lymphocyte-predominant exudate, as in this present case [4], while another study showed polymorphonuclear leucocyte-predominant exudates [5].

The radiological features of actinomycosis differ depending on the primary site of infection and duration of the disease. These features include abscess formation, dense fibrosis, and draining sinuses. Parenchymal features of pulmonary actinomycosis include a peripheral nodule, mass, or consolidation. Therefore, pulmonary actinomycosis can often be misdiagnosed as lung cancer, endobronchial malignancy, or tuberculosis [6]. In the present case, enhancing soft tissue density was found in the left anterior mediastinum on chest CT scan. PET CT was performed under the suspicion of malignancy, and an increased FDG uptake was detected in the lesion. Few reported cases evaluate the appearance of actinomycosis on PET CT scans, and it is universally accepted that a higher SUVmax threshold value than 2.5 indicates malignancy [7]. Therefore, a PET CT scan might not be helpful in differentiating actinomycosis from lung malignancy.

In conclusion, mediastinal actinomycosis is a rare infectious condition and often mimics malignancy. A high level of clinical suspicion is needed to diagnose and treat actinomycosis in patients with indolent, unresolving, or relapsing chronic inflammatory disease. Therefore, in a case of

unexplained exudative pleural effusion, physicians should consider actinomycosis in the differential diagnosis and aggressive tissue confirmation is recommended.

Disclosure Statement

Appropriate written informed consent was obtained for publication of this case report and accompanying images.

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