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CLINICAL CASE REPORT

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Pulmonary Paragonimiasis Mimicking Tuberculous Pleuritis

A Case Report

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Abstract: Pulmonary paragonimiasis is a food-borne zoonosis with a wide variety of radiologic findings, which sometimes can be confused with tuberculosis and carcinoma. Therefore, differential diagnosis is always warranted. A 43-year-old male farmer, with productive cough, blood-tinged sputum and chest pain, as well as patchy consolidation and pleural effusions in chest computer tomography, was misdiagnosed of community-acquired pneumonia and tuberculosis. Complete blood cell count, sputum smear and culture, chest computer tomography, thoracoscopy, and biopsy. The diagnosis of pulmonary paragonimiasis was established due to the finding of Charcot–Leyden crystals in the pleural necrosis, and antibodies against *Paragonimus westermani* in enzyme-linked immunosorbent assay.

Paragonimiasis should be considered as a possibility in the differential diagnosis of tuberculosis. Thoracoscopy is an effective and valuable technology that can help make an accurate diagnosis.

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Abbreviations: ADA = adenosine deaminase, BAL = bronchoalveolar lavage, CBC = complete blood cell count, CEA = carcinoembryonic antigen, CT = computer tomography, ELISA = enzyme-linked immunosorbent assay, Hb = hemoglobin, LDH = lactate dehydrogenase, WBC = white blood cell.

INTRODUCTION

Paragonimiasis, known as pulmonary distomatosis or lung fluke, is a parasitic disease caused by a trematode of the genus Paragonimus via ingestion of raw, inadequately cooked crabs or crayfish.[1] The first report of human lung fluke was in 1879 in China, attributed to the findings of adult worms in the lungs of a dead Taiwanese.[2] The common species of paragonimiasis vary from different regions, with most prevalent species of *Paragonimus westermani* in Asia, whereas *Poroderma africanum* and *Paragonimus uterobilateralis* in Africa.[1] It is estimated that it affects ~22 million people around the world, but with a significant geographical variability of prevalence,[1] resulting in 16.8% in Nigeria,[3] 7% in Liberia,[4] and 1.71% in China.[5]

The onset of paragonimiasis is usually latent due to the chronic course in development.[2] Typical features of pulmonary paragonimiasis are reported to consist of but not limited to cough and blood-tinged sputum in higher frequency, and hemoptysis, distressing chest pain and dyspnea in lower frequency; however, they remained a lack of specificity.[2] Patients with paragonimiasis reveal a wide variety of nonspecific findings in chest radiograph and computed tomography (CT) such as patchy, cloudy infiltration of the lungs, pulmonary nodules, calcified spots, pleural thickening with interlobar pleuritis, pleural effusion, and even masses,[2,6] which are often confused with those in patients with tuberculosis and lung carcinoma.[6,7]

Presenting Symptoms and Clinical Findings

A 43-year-old male farmer was admitted to a local hospital with productive cough, blood-tinged sputum, and chest pain. Complete blood cell count (CBC) showed a neutrophil and eosinophil level of 7510/mm^3 and 270/mm^3, which accounted for 69.9% and 3.1%, respectively. Chest CT showed patchy consolidation in ligule segment of the left lung, atelectasis of the left lower lobe, multiple lymph nodes in the mediastinum, and bilateral pleural effusion. Fluid extracted from the left thoracic cavity was proved to be an exudate with total protein levels of 54.1 g/L, lactate dehydrogenase (LDH) levels of 3262 IU/L, and adenosine deaminase (ADA) levels of 75 IU/L, but the acid-fast staining and the cytological examination of the pleural effusion and sputum were negative. He was diagnosed as community-acquired pneumonia and received antibiotics for a week, and then he was discharged with relief of the symptoms.

Diagnostic and Therapeutic Focus

Two weeks later, the patient was admitted to our hospital for exacerbation of the recurrent chest pain. He had no history of ingestion of freshwater crabs, crayfish, or wild pig meats. On admission, he was in good general health, and the physical examination discovered an enlarged lymph node with a size of ~0.8cm × 1.0 cm in the left anterior triangle and decreased breath sounds in left lower lung with dullness on percussion. After admission, a CBC revealed hemoglobin (Hb) levels of 143 g/L, platelet levels of 256,000/mm^3, absolute white blood cell (WBC) and eosinophil levels of 6620/mm^3 and 390/mm^3, and the percentage of WBC and eosinophil rendering 67.9% and

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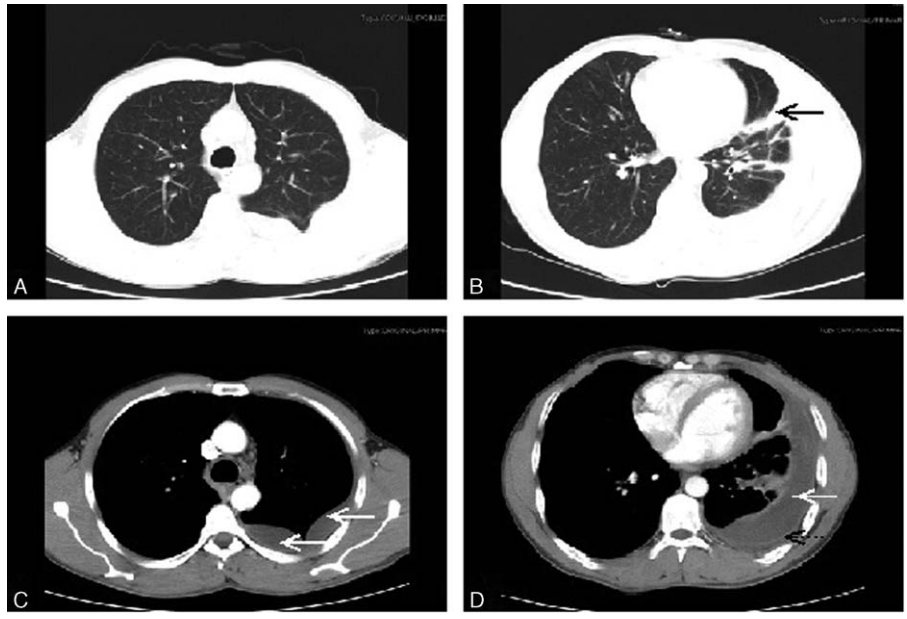

FIGURE 1. Chest CT findings. It showed patchy consolidation in ligule segment of the left lung (black arrow), atelectasis of the left lower lobe, multiple lymph nodes in the mediastinum, left pleural effusion (white arrow), and thickened left pleura (black dashed arrow). CT = computed tomography.

4%. Serum tumor markers showed a normal carcinoembryonic antigen (CEA). Other laboratory tests revealed no abnormal findings. Sputum samples and bronchoalveolar lavage (BAL) found nothing of particular significance. Chest CT showed increased size and positions of patchy consolidation, and thickened left pleura (Figure 1). Thoracoscopy revealed diffused sallow necrosis covered the parietal and visceral pleura (Figure 2), and the biopsy of the necrosis showed chronic inflammation and coagulative necrosis with eosinophils and histocytes infiltration (Figure 3). Thus, tuberculous pleurisy was

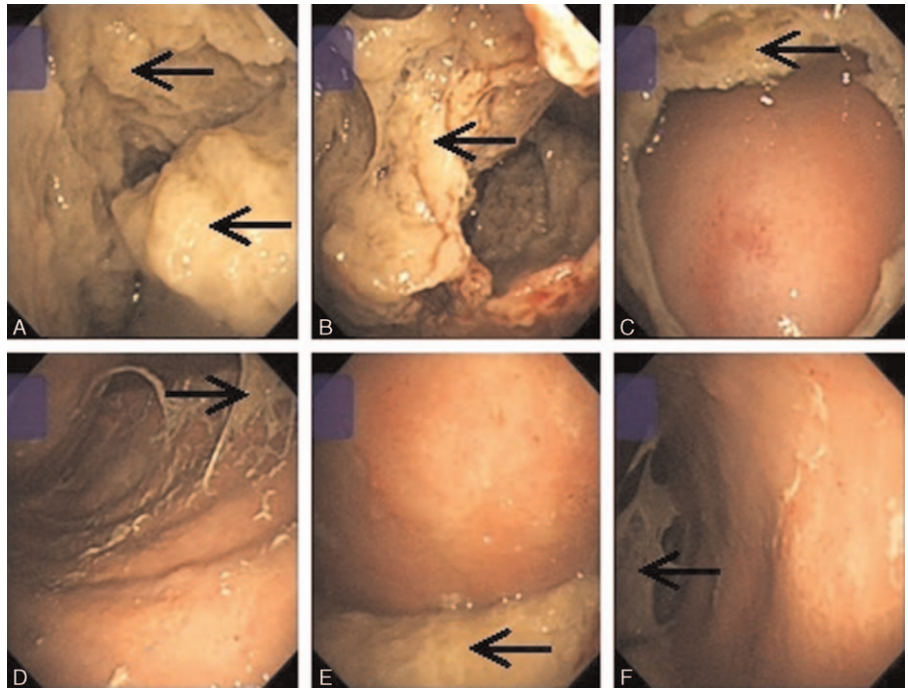

FIGURE 2. Thoracoscopy of left thoracic cavity. It revealed diffused sallow necrosis covered the parietal and visceral pleura (black arrow). (Magnification × 40).

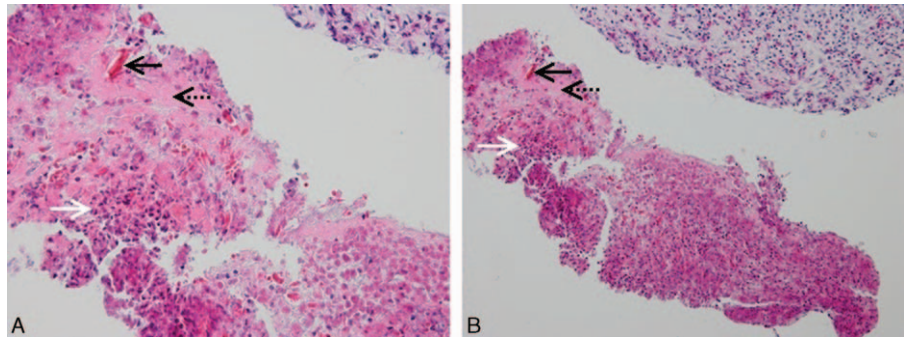

FIGURE 3. Pathological findings of the necrosis in left parietal pleura. They showed chronic inflammation and coagulative necrosis (black dashed arrow) with eosnophils (white arrow) and histocytes infiltration, and Charcot–Leyden crystals (black arrow). (Magnification: 3A × 40, 3B × 10).

suspected and he received diagnostic antituberculosis chemotherapy. For this patient, we could not observe parasite eggs in stool, sputum, or BAL fluid. However, 20 days later, Charcot–Leyden crystals were found in the pleural necrosis (Figure 3), and enzyme-linked immunosorbent assay (ELISA) was positive for antibodies against *P westermani*.

Follow-up and Outcomes

Eventually, this patient was diagnosed as pulmonary paragonimiasis and eosinophilic pleurisy, and he received praziquantel therapy (25 mg/kg, 3 times a day for 3 days). Two months later, he was back to normal life with clear chest CT.

DISCUSSION

Paragonimiasis is a food-borne zoonosis, and it has a parasitic cycle, like all trematode infections, requiring 2 intermediate hosts: first, a mollusc (river snail), where the embryonated eggs become cercariae, and then a freshwater crustacean (crayfish), where they evolve to metacercariae, which are passed to the definitive host (human being or carnivorous mammal) when these crustaceans are ingested in an undercooked state.[1] But sometimes the nonspecificity of the pulmonary symptoms and chest CT may lead to misdiagnosis as pulmonary tuberculosis or lung cancer, as in our patient. In this case with bilateral pleural effusion, thoracentesis of the left thoracic cavity was proven to be an exudate. The most common causes of undiagnosed exudative pleural effusion are tuberculosis and malignancy.[8] But serum tumor markers showed a normal CEA, and thoracoscopy revealed diffused sallow necrosis and the biopsy of the necrosis showed chronic inflammation and coagulative necrosis with eosnophils and histocytes infiltration, so tuberculous pleurisy was suspected and this patient received diagnostic antituberculosis chemotherapy.

Thoracoscopy is defined as the exploration of the pleural cavity and its neighboring organs, and one of its principal applications focuses on the exploration and treatment of pleural effusions.[9,10] Studies demonstrated that thoracoscopy is a safe, easy-to-handle, cost-effective, and accurate technique in diagnosis of pleural effusions of undetermined origin and for obtaining histological diagnosis in pleural effusions.[11] In terms of tuberculosis, the diagnostic sensitivity ranges from 93.3% in areas of low prevalence for tuberculosis to 100% in areas of high prevalence.[12,13] Case report of pulmonary paragonimiasis diagnosed by thoracoscopy showed a dense thickened peel over both visceral and parietal pleural surfaces, and the histopathological examination of pleural biopsy demonstrated that eosinophilic pleuritis with polarizable birefringent material was in the center of oval-shaped necrotizing granulomas and that the lung parenchyma was heavily infiltrated with macrophages and eosinophils.[14] Although in the case of tuberculous pleurisy, thoracoscopy showed that diffuse nodules on pleura and biopsy specimen demonstrated epithelioid cell granulomas with caseation necrosis,[15] in the present case, thoracoscopy revealed that diffused sallow necrosis covered the parietal and visceral pleura, and the biopsy specimen showed chronic inflammation and coagulative necrosis with eosinophils and histiocytes infiltration, which corresponded to both conditions, and tuberculous pleurisy could not be excluded.

However, our patient was finally diagnosed as pulmonary paragonimiasis because Charcot–Leyden crystals were found in the pleural necrosis and ELISA was positive for antibodies against *P westermani*. Charcot–Leyden crystals are bipyramidal crystals seen in body fluids or aspirates associated with eosinophilic inflammatory reactions.[16] They are composed of lysophospholipase, a constituent protein of eosinophils, and are formed when increased turnover of eosinophils occurs, and it is postulated that they degrade the lipases generated on cell necrosis.[17] In addition, they can also be occasionally seen in patients with other diseases besides paragonimiasis,[18] such as acute myeloid leukemia,[19] invasive aspergillosis[20] among others, which, however, could easily be differentiated from paragonimiasis based on the clinical presentations and related laboratory analysis. Eventually, in combination of positive ELISA test for paragonimiasis, pulmonary paragonimiasis was identified.

As a possibility in the differential diagnosis of increased serum eosinophils and exudative pleural effusion with thickened pleura, paragonimiasis should be considered. Thoracoscopy is an effective and valuable technology that can help make an accurate diagnosis.

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