Case Report

Pulmonary Paragonimiasis: A Great Masquerader

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Abstract

Pulmonary paragonimiasis is one of the rare causes of haemoptysis in humans. The clinical findings of paragonimiasis may mimic pleuro-pulmonary tuberculosis (TB), bronchitis, bronchiectasis, pneumonia, epilepsy or cerebral space-occupying lesion. It mimics pulmonary TB due to overlapping symptoms and it requires differentiation from the same in TB endemic areas. We carried out retrospective analysis of four cases of pleuro-pulmonary paragonimiasis who presented to our centre during the period from 2013 to 2017. All four cases had presented with streaky haemoptysis, chest pain and on evaluation were found to have pulmonary eosinophilia. One patient also had abdominal manifestations. Paragonimus eggs were demonstrated in respiratory secretions of three cases while one had eosinophilic pleuritis. Failure to suspect pulmonary paragonimiasis at initial evaluation results in wrong diagnosis as it masquerades many other diseases with common symptomatology.
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Key words: Pulmonary eosinophilia, *Paragonimus westermanii*, Endemic haemoptysis.

Introduction

Paragonimiasis has been an important food-borne parasitic zoonosis. It is caused by trematode of the genus *Paragonimus*.[1] It has been recognised as an important cause of pulmonary disease worldwide, especially in Asia, West-Central Africa, and Central and South America.[2] The clinical findings of paragonimiasis may mimic pleuro-pulmonary tuberculosis (TB), bronchitis, bronchiectasis, pneumonia, epilepsy or cerebral space-occupying lesion. It mimics pulmonary TB and requires differentiation from the same in TB endemic areas. The signs and symptoms of initial presentation are most commonly haemoptysis, pleural effusion and peripheral blood eosinophilia. Diagnosis may be missed, especially in areas not endemic for the parasite.[2] Approximately 50% to 70% of patients on initial work-up are diagnosed and treated as TB. The country of origin and dietary habit of eating raw, or partially cooked crab or crayfish are important features in a patient suspected to have paragonimiasis.[3] Hence, a high index of suspicion should be kept in mind if one comes across a patient from an endemic area who has predominantly lower respiratory tract symptoms, including persons with normal chest radiograph and those with radiograph suggestive of TB.

Case Reports

Case 1

A 34-year-old male, resident of Nagaland presented with complaints of fever, breathlessness. Physical examination revealed splenomegaly and bilateral pleural effusion. He had raised total leukocyte count with 70% eosinophilia. Absolute eosinophil count (AEC) was 11,000/mm^3. Chest radiograph revealed bilateral pleural effusion; diagnostic testing showed that the pleural effusion was exudative and had eosinophilia. Ultrasonography of abdomen showed mild splenomegaly and mild ascites. Bone marrow biopsy revealed cellular bone marrow with prominence of eosinophilia. Testing for FIP1L1 platelet-derived growth factor receptor alpha (F1P1L1-PDGFRα) gene rearrangement was negative. Patient was initially managed as a case of hyper-eosinophilic syndrome and was treated with a course of diethyl-carbamazine and oral corticosteroids along with pegylated interferon (PEG IFN) by haematologist. However, he again came after two months with complaints of fever, dry cough and streaky haemoptysis. At this time history of eating raw and pickled crustaceans was elicited. Computed tomography (CT) of chest (Figure 1) showed bilateral nodular opacities with ring cyst appearance.

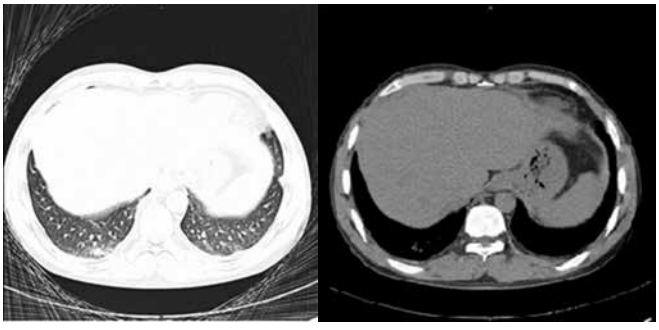

Figure 1. Computed tomography of chest showing bilateral reticulo-nodular opacities with ring cyst appearance in Case 1.

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Case 2

A 28-year-old male, resident of Nagaland, old treated case of pulmonary TB had presented with complaints of pain abdomen, diarrhoea and streaky haemoptysis. Patient had a history of consumption of pickled crab. Laboratory evaluation revealed leucocytosis with eosinophilia (65% to 70%); AEC was 17,000/mm^3. Peripheral blood smear showed no microfilariae. Chest radiograph revealed few scattered bilateral interstitial opacities. Ultrasonography of the abdomen was normal. Serum immunoglobulin E (IgE) levels were raised. Upper gastrointestinal endoscopy (UGIE) showed mild gastroduodenitis. CT of the chest (Figure 2) showed bilateral reticulo-nodular opacities with ring cyst appearance. F1P1L1-PDGFRA gene re-arrangement was not seen.

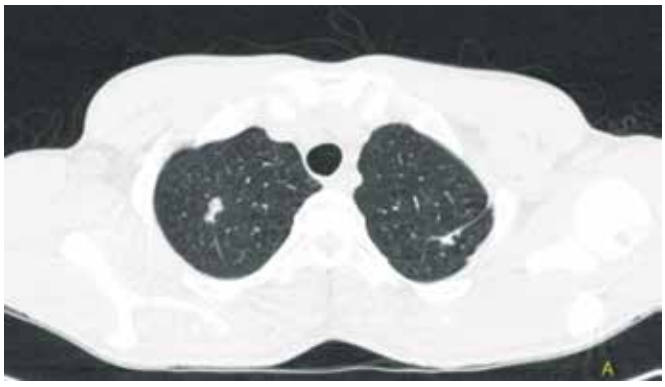

Figure 2. Computed tomography of chest showing bilateral reticulo-nodular opacities with burrow track in Case 2.

Case 3

A 39-year-old male, resident of Nagaland presented with complaints of left-sided pleuritic chest pain, dry cough, breathlessness and weight loss. He was already receiving anti-TB treatment in view of clinical suspicion. Leucocyte count was 9200/mm^3 with eosinophilia of 20%. AEC was 1740/mm^3. Peripheral blood smear had shown no evidence of atypical cells. Stool examination was normal. His total serum immunoglobulin E (IgE) was 3100 IU/mL and chest radiograph revealed left-sided pleural effusion. Pleural fluid analysis revealed exudative pleural effusion with pleural fluid glucose 28mg/dL, lactate dehydrogenase (LDH) 3500IU/L and eosinophilia. Pleural fluid adenosine deaminase was 58U/L. Chest CT revealed bilateral pleural effusion. Retrospectively patient gave history of eating raw and pickled crabs. Patient underwent medical thoracoscopy for pleural effusion.

Case 4

A 32-year-old male, resident of Nagaland presented with complaints of cough with expectoration, fever, pleuritic chest pain and streaky haemoptysis. He gave history of eating raw crustaceans. Total leucocyte count was 10100/mm^3 with 30% eosinophilia. AEC was 2730/mm^3. CT of the chest revealed peripheral consolidation in superior lingular segment of the left upper lobe. Stool examination had no evidence of ova or cysts. Peripheral blood smear had no atypical cells or parasites. Bone marrow study was essentially normal except eosinophilia. FIP1L1-PDGFR α mutation study revealed no abnormality. Bronchoalveolar lavage cytology showed 13% eosinophils.

Microscopic examination of respiratory samples was done in all the cases. Consecutive sputum samples were examined in Case 2 while bronchoalveolar lavage (BAL) sample examination done in Case 1 and 4. Case 3 underwent medical thoracoscopy. BAL sample had a brownish supernatant in case 1. Sputum and BAL sample in case 1, 2 and 4 on iodine mount revealed operculated cysts of *Paragonimus westermani* (Figure 3). While in case 3 pleural fluid was exudative with low glucose, high lactate dehydrogenase (LDH) and eosinophilia. Thoracoscopic pleural biopsy revealed eosinophilic pleuritis. All patient were managed with praziquantel 25mg/kg body weight three times daily for three days. They responded well with resolution of clinical symptoms. Follow-up evaluation revealed resolution of radiological findings and normal haemogram.

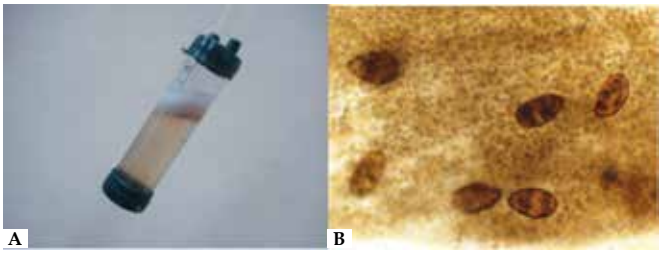

Figure 3. (A) Bronchoalveolar lavage specimen showing brownish supernatant and (B) photomicrograph showing ova of paragonimus species on iodine mount (magnification x 400).

Discussion

Paragonimiasis, also known as endemic haemoptysis is caused by the trematode species of the genus *Paragonimus*.[1] Human are mammalian definitive hosts of this parasite. Crabs, fresh water snails and crayfish are among the intermediate hosts of the parasite. Humans acquire the infection most commonly by ingestion of uncooked or undercooked crustaceans[4] and eliciting dietary history is of paramount importance. Paragonimiasis may be classified on clinical context into pulmonary, extra-pulmonary and pleuro-pulmonary forms. All our cases had pleuro-pulmonary manifestation except Case 2 who presented with abdominal symptoms and had pulmonary involvement radiologically. It can also be classified into acute, chronic and ectopic form of paragonimiasis.[5] Initial presentation can be with complaints of chest pain, difficult breathing, and coughing up rusty brown or blood-stained sputum or recurrent haemoptysis. All our patients presented with constitutional symptoms associated with episodes of haemoptysis except one who had pain abdomen at

presentation. Migrating worms in the pleural cavity can also produce pleuritis and pleural effusion.[1,5,6] Two of our patients had eosinophilic pleural effusion. Case 2 had pain abdomen which can be attributed to gastrointestinal penetration.[5]

Definitive diagnosis of the disease can be made by demonstrating characteristic ellipsoid, golden brown, operculated *Paragonimus* eggs in the respiratory tract secretions as in our case series, however it is not usually seen in the pleural effusion. Peripheral eosinophilia in blood smear is a supporting evidence which was present in all our cases. The main chest computed tomography feature of pleuro-pulmonary paragonimiasis is pulmonary nodules. These are also known as worm nodule connected to the pleura by a linear track. Sub-pleural streaky opacity connecting the pleura and the nodule are presumed to be a worm migration track (burrow track).[7] Three of our cases had radiological findings of nodular opacities with burrow track, ring cyst opacities, pleural effusion and consolidation. Pleural effusions appear to be more common in early stages of the infection.[8] One of our cases had eosinophilic pleural effusion with low glucose, high LDH and with eosinophilic pleuritis on histopathology. Pleural fluid findings of low glucose, high LDH, high protein and low pH in the presence of eosinophilia with compatible clinical, radiologic findings allow the diagnosis even in the absence of demonstrable ova in pleural fluid.[9] Praziquantel at a dose of 75mg/kg per day for three days is the drug of choice for paragonimiasis[10] and all our patients responded well to it.

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