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Unilateral Massive Pleural Effusion Mimicking Pulmonary Tuberculosis Due To Pleuropulmonary Paragonimiasis- A Case from Manipur, India

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Abstract: *Paragonimiasis is a food-borne zoonotic disease caused by the genus Paragonimus which usually results from the intake of uncooked or undercooked freshwater crab or crayfish. Manipur is a northeastern state of India which is endemic for paragonimiasis where the first Indo-Japan joint research on Paragonimus and paragonimiasis was conducted to study the life cycle, pathobiology, and morphological characterization of Paragonimus species in India. Diagnosis of paragonimiasis is often difficult due to lack of awareness and atypical presentation which usually mimic other chronic pulmonary illness such as tuberculosis. Diagnosis of paragonimiasis is by detection of ova in the sputum,pleural fluid or tissue specimen or serology. The disease can be successfully treated by praziquantel. Here we report a case of pulmonary paragonimiasis presented as right sided massive eosinophilic pleural effusion from an area endemic for paragonimiasis which was initially mistaken as smear negative pulmonary tuberculosis.*

Keywords: *Pleural effusion, Peuropulmonary, Paragonimiasis.*

I. Introduction

Paragonimiasis is a food-borne zoonotic disease caused by trematode worms of the genus Paragonimus commonly Paragonimus westermani (popularly known as oriental lung fluke) , which is also known as endemic haemoptysis, oriental lung fluke infection, pulmonary distomiasis, parasitical haemoptysis, parasitarehaemopte, Gregarinosispulmonum, etc.[1]Paragonimus species are endemic to Southeast Asia, Latin America (most commonly in Peru), and Africa.[1]No record of scientific research on Paragonimus and paragonimiasis in India was available until late 1900s till the first epidemiological survey of paragonimiasis in India was conducted in Imphal east district, Manipur during the period from 1986 and 1987.The first Indo-Japan joint research on Paragonimus and paragonimiasis was conducted in Manipur in 1990 to study about the details of of Paragonimusspecies. This resulted in the discovery of endemic areas of paragonimiasis, identification of intermediate hosts and morphological characterization of Paragonimusspecies occurring in Manipur.[1,2] Eventhough P. westermani has been regarded as the most common pathogen, P. heterotremus has been increasingly detected as an important cause of infection in humansin South and Southeast Asia.[1,3] Humans acquire lung fluke infection by ingesting infective metacercariae encysted in the muscles and viscera of crayfish and freshwater crabs. Prevalence of infection in endemic areas ranges from 0.1-23.75% and it is 6.7% in Manipur according to Singh TS et al.[2] Paragonimiasis cases have been detected almost every year in Manipur but only a few have been published. In this case,the patient presented as right sided massive eosinophilic pleural effusion from an area endemic for paragonimiasis which was initially mistaken as smear negative pulmonary tuberculosis ultimately diagnosed as paragonimiasis.

II. Case report

25 year old male from Sugnu, Churchandpur district which is a remote village of Manipur was suffering from chest pain for the past one year on and off. It was occasionally associated with shortness of breath, non productive cough and haemoptysis. Inspite of multiple courses of antibiotic therapy symptoms persisted. Local treating physician suspected pulmonary tuberculosis but sputum smear was negative for acid fast bacilli and antitubercular treatment was started as he suspected smear negative pulmonary tuberculosis. Mantaux test was also negative. Antitubercular treatment was given for 6 months, but symptoms reappeared after a short period of temporary relief. Ultimately the patient was referred to Cardiothoracic OPD of Regional Institute of Medical Sciences which is a tertiary care hospital of the state. Right sided pleural effusion was diagnosed clinically. Examination of other systems were within normal limits. On routine work up we noted absolute eosinophilia with otherwise normal haemogram. Liver and kidney function tests were within normal limits. X ray chest showed massive right sided pleural effusion(Fig.1). CT scan of thorax confirmed right sided pleural effusion with passive atelectasis of adjecent lung(Fig.2). Tube thoracostomy was done as an immediate

treatment. Patient was negative for HIV, hepatitis B and C. Pleural fluid analysis showed mixed inflammatory cells which predominantly composed of eosinophils. Gram staining of pleural fluid showed pus cells but no organisms were detected. Routine bacteriological culture and AFB staining were negative. Paragonimiasis was suspected since it is a case of eosinophilic pleural effusion. To our surprise patient gave the history of regular intake cooked as well as raw crabs. Serologic test for paragonimus came as positive. No egg or fluke was detected in the pleural fluid. Praziquantal 75 mg/kg/day was given for 3 days. Regular chest exercises was advised. Thoracotomy was done because of suspected pleural thickening and residual collection in the pleural cavity which was hindering the lung expansion. Decortication with the removal of necrotic material was done and approximately about 500 grams of putty like yellowish white necrotic material was removed intraoperatively(Fig.5&6). Histopathology report didn't show any evidence of malignancy, tuberculosis or other chronic illness. General condition of the patient was improved and he was discharged after 10 days of surgery without any post operative complications.

III. Discussion

Pulmonary paragonimiasis is a subacute to chronic inflammatory disease of the lung caused by trematodes of the Paragonimus genus most commonly oriental lung fluke, Paragonimus westermani. The disease primarily affects respiratory system. Other systems affected are central nervous system, gastrointestinal system etc. Flukes may also be found in the liver, spleen, peritoneum, intestinal wall, and intraabdominal lymph nodes. The adult trematode is reddish-brown and ovoid and the eggs are golden brown asymmetrically ovoid, have a thick shell with an operculum. When humans ingest raw infected crustaceans, larval flukes develop in the small intestine and penetrate the intestinal wall into the peritoneal cavity after excysting. They then migrate into the abdominal wall or liver, where they undergo further development. Later, adult flukes reenter from the abdominal cavity and penetrate the diaphragm to reach the pleural space and lungs. The eggs may then be expectorated or swallowed. If these eggs reach a water source, the life cycle will start over again. Flukes mature, a fibrous cyst wall develops around them, and then egg deposition starts 5-6 weeks after infection.[1,4] Death may occur during the acute phase of infection. For those who survive the acute phase, spontaneous recovery usually occurs within 1-2 months, but symptoms may recur intermittently over several years. Paragonimiasis are asymptomatic in 20% of cases. Abdominal pain, diarrhea, and urticaria occur during the acute phase, which corresponds to the period of invasion and migration of immature flukes. These initial symptoms are followed a few days later by fever, cough, dyspnea, chest pain, malaise, and sweats. The acute phase usually persists for several weeks. During the chronic phase, manifestations may be pulmonary or extrapulmonary. Chronic pulmonary symptoms consist ofdry cough followed by a cough productive of tenacious and rusty or golden sputum. Pulmonary symptoms begin approximately 6 months after infection and are often mistaken for symptoms of tuberculosis (TB). Eosinophilia and lack of fever suggest the true diagnosis. Patients frequently report vague chest discomfort, dyspnea on exertion, or wheezing. Life-threatening hemoptysis may occur in some cases.. Radiographic abnormalities include pleural effusions, air space consolidation, linear opacities, thinwalled cysts, mass lesions, nodules or pleural thickening.[4,5]Extrapulmonaryparagonimiasis can be divided into cerebral, abdominal, subcutaneous, and miscellaneous forms of the disease.Definitive diagnosis of paragonimiasis requires detection of eggs in sputum, feces, pleural fluid, cerebrospinal fluid (CSF), or pus.[4]

Paragonimiasis usually misdiagnosed as tuberculosis.[1,4,6,7] Sharma DC et al.[6]described in a study from India that 60% of paragonimiasis cases were confused with smear negative tuberculosis.Singh TN et al.[7] reported three cases with clinical and radiological features compatible with pulmonary tuberculosis which were negative for sputum acid fast bacilli and previously treated as smear negative pulmonary tuberculosis. Symptoms persisted inspite of antitubercular treatment and all cases were ultimately diagnosed as pleuropulmonary paragonimiasis.

Pleuritis and effusion is produced due to the migrating worms in the pleural cavity. Severity of effusion is depending upon the number of worms, frequency of migration and duration. Progressive dyspnoea, chest pain and cough with or without haemoptysis are the main presenting symptoms of patients with pleural effusion.[1] According to Uchiyama et al.,[8] 35 % of pleural effusions were unilateral and 5% were bilateral. Vidamaly et al.[4] reported nine cases of persistent pleural effusion in whom 44.4% patients showed bilateral pleural effusion due to paragonimiasis in Lao PDR and they suggested that paragonimiasis should be considered in any case of elusive pleural effusion occurring in endemic areas. Authors also share the same opinion from our experience. Eventhough paragonimiasis usually affects those who consume raw or undercooked crab,crayfish or sliced meat of wild boar, it can rarely affects those who didn't cosume these also. Mizuki et al.[9] reported a case of paragonimiasis with pleural effusion eight months after migrating subcutaneous induration of the abdominal wall in a patient who had never eaten fresh water crabs or meat of wild boar.

IV. Conclusion

Paragonimiasis usually misdiagnosed as tuberculosis. It should be considered as a differential diagnosis in any case of long standing pleural effusionoccurring in endemic areas.

Acknowledgement

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Fig.1 and 2 :-Chest X ray and chest CT scan at the time of presentation showing massive right sided pleural effusion respectively.

Fig. 1

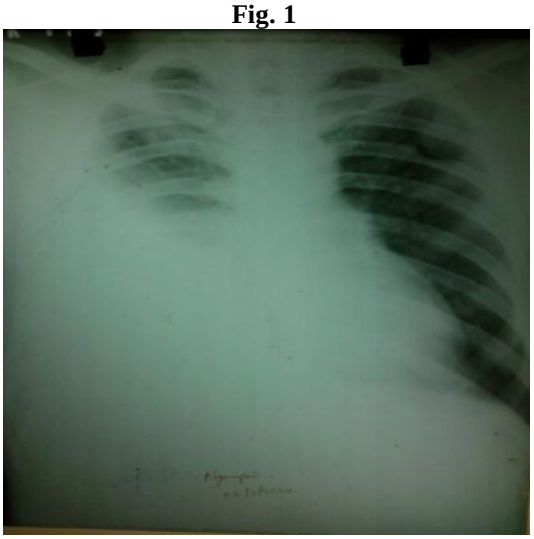

fig. 2

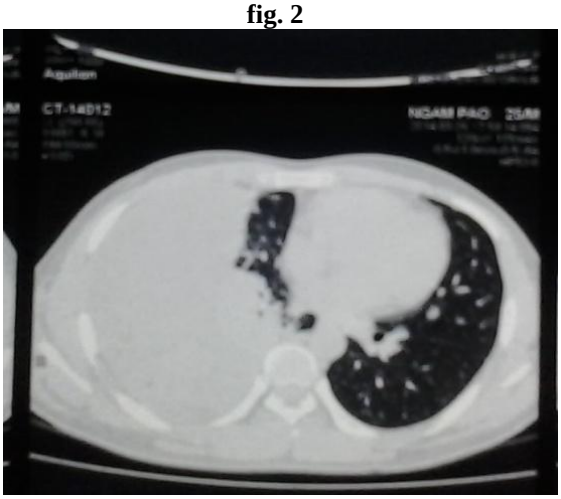

Fig. 3 and 4:- Chest X ray before and after thoracotomy

Fig.3

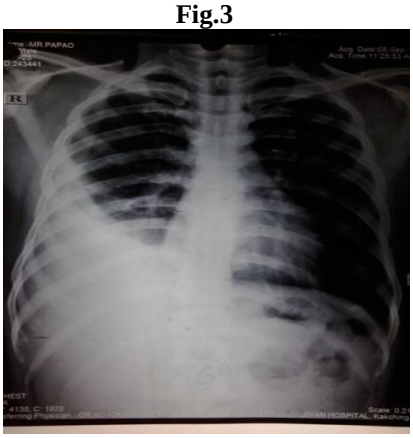

Fig.4

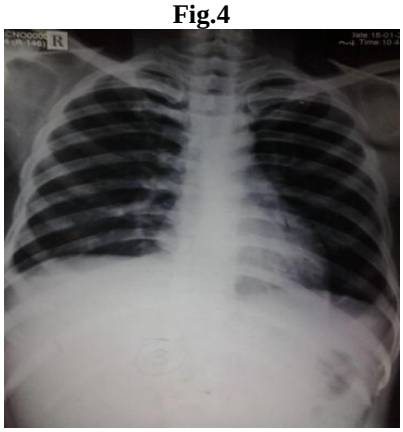

Fig 5 and 6 showing putty like inflammatory material which was removed from the pleural cavity during operation.

Fig. 5

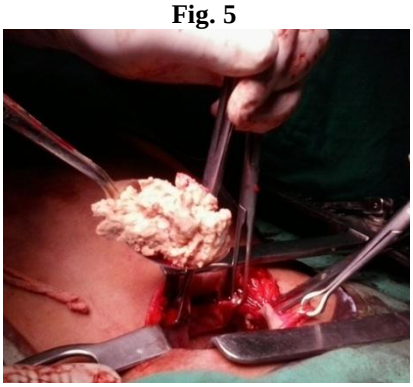

Fig. 6