A REPORTED CASE OF PULMONARY SPARGANOSIS : RADIOLOGICAL ASPECT AND REVIEW OF ARTICLES.

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ABSTRACT

Human sparganosis is a rare parasitic infestation caused by the migratory second stage larvae (plerocercoid) of the Pseudophyllidean cestode of genus Spirometra (Mueller, 1973). The first report of human sparganosis was made by Patrick Manson in a Chinese patient in 1882 (Beaver, 1984). The distribution of the cases was sporadically worldwide, but most commonly from East and Southeast Asia (Japan,China,Korea, Taiwan, Vietnam and Thailand). There were some reports from the Southern part of the United States and Europe (Mueller et al, 1975). Sparganosis was commonly reported to involve subcutaneous tissue, muscle, ocular and visceral organs including the central nervous system. Pulmonary involvement is extremely rare. A case of pulmonary sparganosis was presented with chronic hemoptysis and abnormal lung shadows. The diagnosis could not be made by radiological image and bronchoscopic examination.The patient received an antituberculous drug without response. Finally the diagnosis was confirmed by open lung biopsy which the plerocercoid larvae of sparganum was found.

INTRODUCTION

Sparganosis is an uncommon zoonotic disease caused by infection with Pseudophyllidean cestodes of the genus Spirometra. There are two types of the disease, namely nonproliferative and proliferative sparganosis.

Human infection occurs by one of the three routes:ingestion of freshwater cyclops which harbor an infective procercoid; ingestion of the plerocercoid directly from an intermediate animal hosts such as frogs or snakes ; or by percutaneous infection from crude poultices prepared from infected amphibians or reptiles. Once inside the host, larvae can migrate to various tissues, usually encysts in subcutaneous tissue, ocular involvement is not uncommon including the central nervous system. Pulmonary involvement is extremely rare. We report a case of pulmonary sparganosis which was misdiagnosed and treated as tuberculous infection consequently there was no improvement in the patient. Subsequently surgical open lung biopsy resulted in confirmation of the diagnosis.

CASE REPORT

A 37-year-old Thai man lived in Banpur district, Udonthani province (Northeastern part of Thailand). The patient presented with history of chronic cough, hemoptysis for about 3 months with low grade fever. He had a history of habitually consuming impure water but denied a history of having eaten raw or partially cooked meat. Seven years ago he contracted spinal tuberculosis and was treated for 1 year. Ten years

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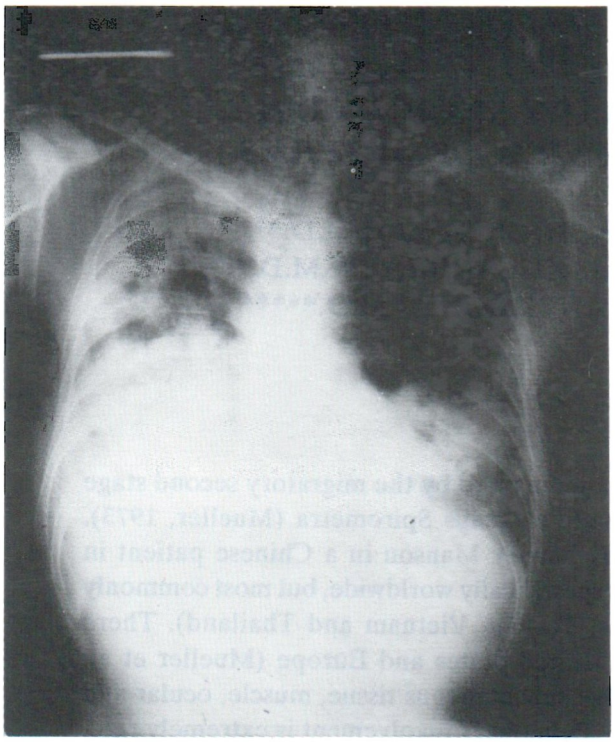

Fig. 1a PA chest film shows diffuse bilateral patchy alveolar infiltration with some cystic lesions within. More confluent lesions are noted on the right lung with bilateral pleural fluid.

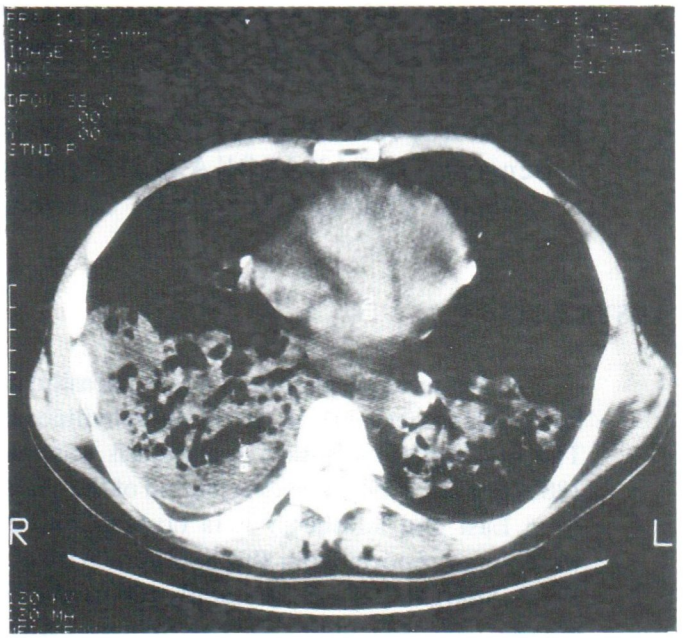

Fig. 1b

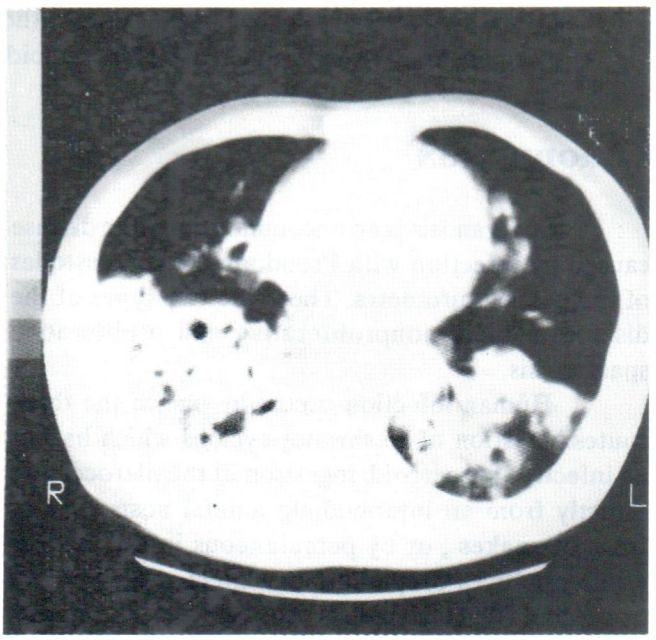

Fig. 1c

Fig. 1b, and 1c Computed tomography of the chest clarifies abnormal lung shadows as areas of consolidation with multiple thick wall cavitary lesions and tubular bronchiectatic changes.

ago he had an operation on the right kidney due to a renal stone. Physical examination revealed a middle aged man with mild thoracic kyphosis. Neither evidence of organomegaly nor lymphadenopathy was detected. Chest examination revealed fine crepitation in both lungs. Laboratory examination revealed a hematocrit of 36% and a white blood count of 11200/ mm^3 with a differential of polymorphonuclears 60%, lymphocytes 23%, monocytes 6% and eosinophils 10%. Pulmonary function showed a restrictive pattern.

Roentgen examination including chest x-ray revealed diffuse bilateral patchy alveolar infiltration more confluent on the right lung with some cystic cavitary lesions, suspected to be due to the pulmonary infection process.

Bronchoscopic examination revealed no endobronchial lesion and mild inflammation of the mucosa with contact bleeding. Cytologic examination revealed a negative AFB stain and unremarkable findings from bronchial biopsy.

The patient was treated for 2 months as having tuberculosis without improvement. He was lost to follow

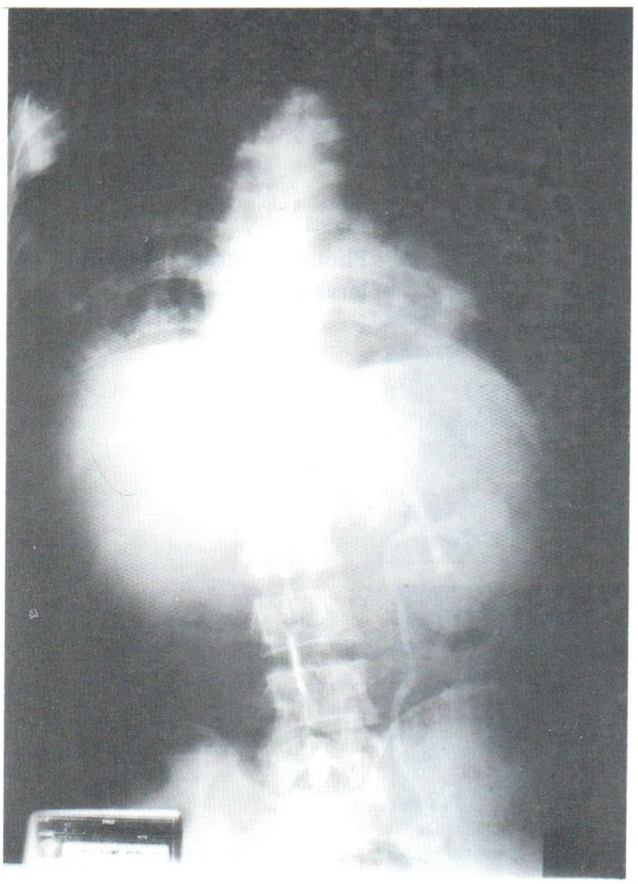

Fig. 2 Plain KUB following computed tomographic examination reveals excretion of contrast media only on the left kidney.

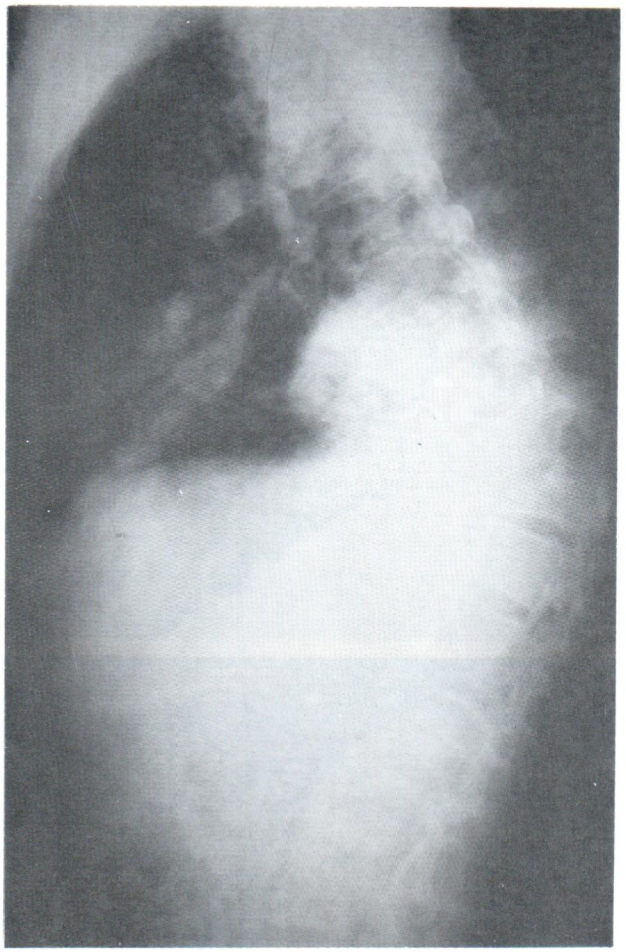

fig. 3 Lateral thoracic spine shows anterior wedging of T12 vertebral body.

up for 8 months and then came back again with clinical signs of chronic productive cough. A chest film still noted abnormal diffuse patchy alveolar shadows, which were well delineated on additional computed tomography. An area of lung consolidation with air bronchogram, multiple thick wall cavitary lesions right side and tubular bronchiectatic changes were also noted in both lower lungs with bilateral pleural effusion more on the right side. (fig 1a, 1b, 1c). The findings were suggestive of a pulmonary infection process. Plain film of the spine showed anterior wedging and collapse of T12 body with mild narrowing of intervertebral disc space of T11-12 level. We also noted expansile permeative destruction of the mid part of the posterior 11^{th} rib and upper cortical destruction of 12^{th} rib on the right side.

Repeated bronchoscopic examination was done which revealed mild inflammation of the mucosa but the cytologic examination was normal. He received a short course of antibiotics with minimal improvement of the symptoms. Finally open lung biopsy was carried out. Sections of the lung showed multiple foci of sparganum encyst in the lung tissue. The cystic space was lined by fibrous connective tissue, which was infiltrated by eosinophils,plasma cells and lymphocytes. Scattered macrophages contained hemosiderin pigment. Foci of a necrosed parasite with calcification were seen. Other areas of lung tissue showed atelectasis with hemorrhage. (fig 4,5)

The patient received Mebendazole 40 mg/kg/d and was followed up for about 5 months with minimal weight gain and slight improvement of clinical symptoms.

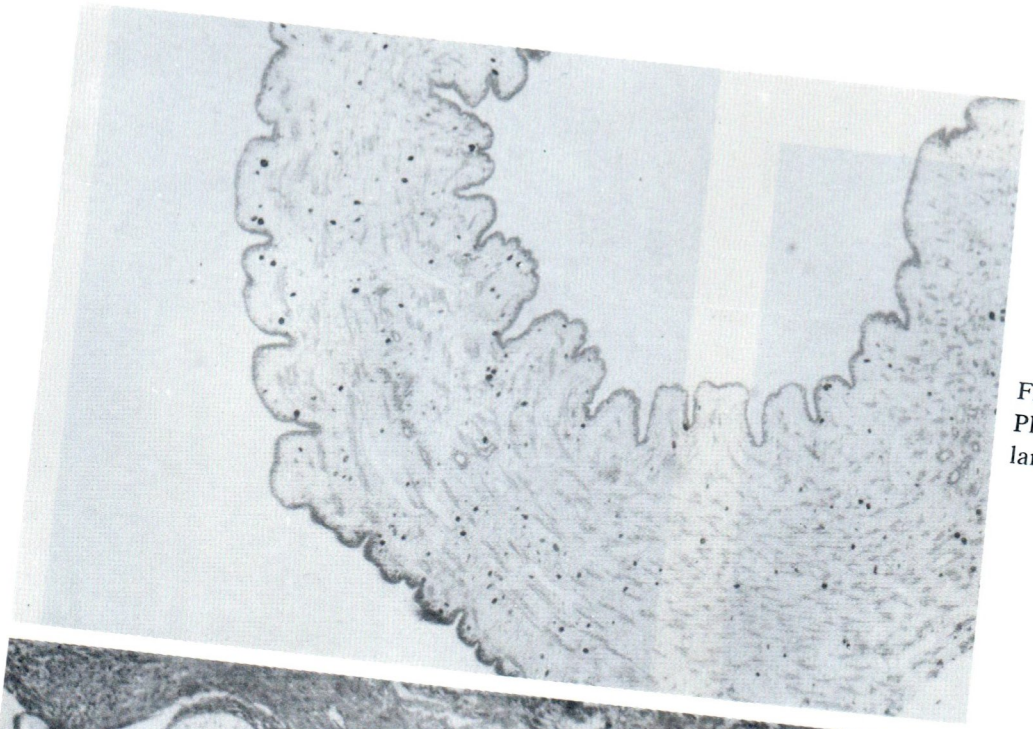

Fig. 4
Photomicrograph of sparganum larva (33x)

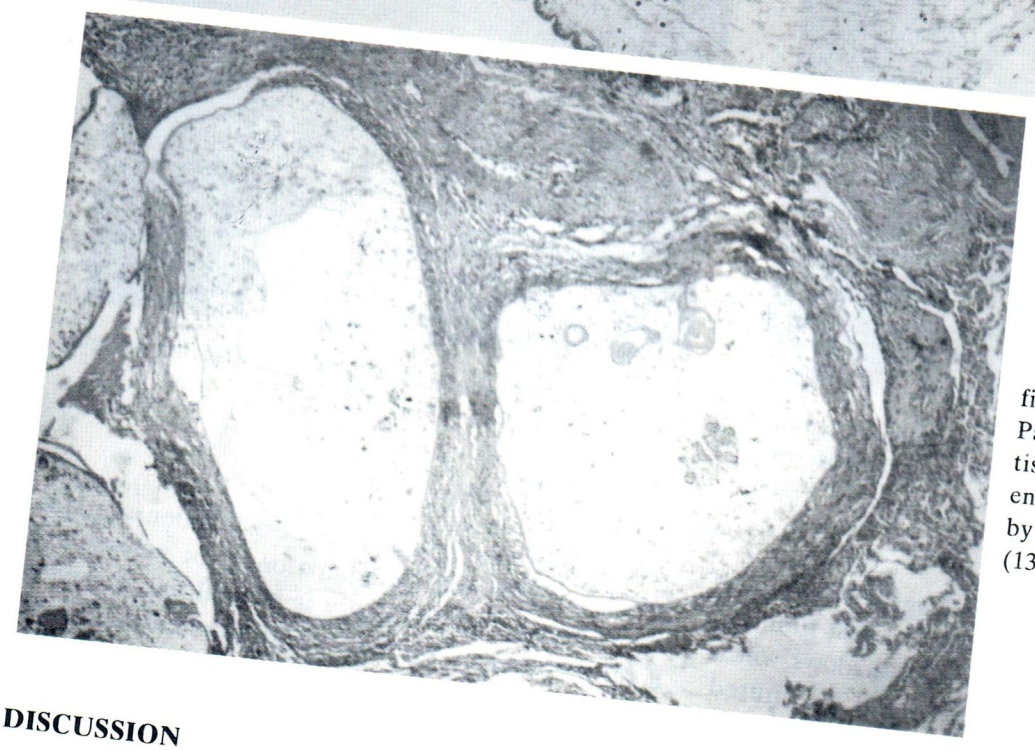

fig.5
Pathologic section of the lung tissue shows multiple foci of encysted sparganum surrounded by inflammatory cell reaction (13.2x)

DISCUSSION

Sparganosis is an uncommon parasitic infestation of man caused by larval tapeworms[2]. The eggs of the adult worms hatch in water and develop into larvae in Cyclops species, which are swallowed by intermediate hosts, such as frogs, snakes and lizards. Larvae develop and are finally ingested by the definitive host (dogs and cats). Humans become accidentally infected. Humans can be infected by three routes[12]. The first is by drinking unfiltered water contaminated with infected cyclops. The second is by ingestion of the second intermediate host such as raw or inadequately cooked flesh of snakes or frogs which contains living larvae (the most common source of human infection in Korea)[10]. The third route is by applying the flesh of an infected intermediate host to an open wound or sore eyes as "poultices", which is practised in China and Southeast Asian countries[1,3,6]. The infection route in this patient is unknown, but most likely, it is by drinking contaminated water.

When humans are infected by first or second

stage larvae, the larvae cannot develop into adult worms and will live in the tissues in the "sparganum" stage. Some authors believed that the larvae could live in the tissue for more than 20 years[4,5]. While living in the human hosts, the larvae can migrate, causing symptoms such as migratory swelling in subcutaneous tissue. They can lodge in tissue producing mass lesions or penetrate the gastrointestinal wall to migrate systemically. Pulmonary sparganosis is extremely rare[8]. The diagnosis in this patient can not be made preoperatively. The investigation showed eosinophilia, although serology and radiological evaluation is helpful, the definite diagnosis requires direct identification of the parasite from surgical specimens[5]. The characteristic pathologic findings of sparganosis is focal necrosis along the tortuous tract of migration. The track is surrounded by a zone of necrotic debries, lymphohistiocytic reaction and heavy eosinophilic infiltration are usually found. The inflammatory reaction often extends into surrounding tissue, particularly during the acute migration phase. Pulmonary pathology in this case can explain radiographic images retrospectively.

An aspect of prevention of this disease, should be the education of the community who live nearby in the endemic area about hygiene and public health especially in the field of sparganum life cycle.

ACKNOWLEDGEMENT

We are grateful to Mr.E.W. Renton for reviewing the manuscript.

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