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HELMINTHOLOGY - SHORT COMMUNICATION

First report of sparganosis manifested by pleuritis and decreased peripheral blood eosinophils in Jiangsu province, China

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Abstract

Sparganosis is a parasitic infection caused by the metacestode stage of *Spirometra mansoni* and some other related diphyllobothriidean cestodes. Although various internal organs were involved in *sparganum* infection, pulmonary and pleural involvement is rarely reported. We herein report an uncommon form of sparganosis manifested by pleuritis and decreased peripheral blood eosinophils. *Sparganum* worms were found in the pleural effusion accidentally and confirmed by pathological diagnosis. After being treated with praziquantel for 10 days, the patient's symptoms, laboratory examinations, and imaging findings were improved gradually.

Keywords *Spirometra mansoni* · Sparganosis · Pleural effusion · P. R. China

Introduction

Sparganosis is a parasitic infection caused by the plerocercoid metacestode of *Spirometra mansoni* and related diphyllobothriidean cestodes. It has been reported sporadically around the world, especially in Asian countries, including Korea, Japan, Thailand, and China (Fukushima and Yamane 1999). Humans are generally infected through eating raw or inadequately cooked frogs and snakes or drinking unboiled stream water (Walker and Zunt 2005). Some aboriginal inhabitants get infected through placing frogs' or snakes' flesh on open wounds, which is considered as a traditional treatment of skin ulcers or eye disease (Chen et al. 2015). Additionally, eating live tadpoles is another route of plerocercoid infection for humans in some areas of China (Cui et al. 2011a, b). The plerocercoid migrates to soft subcutaneous tissues or muscles and further invades the brain, spinal cord, eyes, intestines, urogenital organs, pericardium, or lymphoglandula (Cho et al. 1975). Patients with sparganosis usually present eosinophilia and elevated IgE levels (Ishii et al. 2001). Here, a rare case of sparganosis characterized by pleural effusion and decreased peripheral blood eosinophils is reported.

Case report

A 58-year-old man was admitted to our hospital with left chest pain and shortness of breath for 1 week. He also presented fever, cough, expectoration, and gradually felt dyspneic while in the supine position. The dyspnea was abated via sitting up or changing into the lateral position. The patient had a history of asthma for 54 years. He has been treated with prednisone for a long duration and inhaling ventolin discontinuously. His personal history included social drinking and smoking occasionally, without a history of eating raw frogs or snakes.

He presented with an acute ill-looking appearance and a body temperature of 37.8 °C. The initial laboratory test results showed an increased white blood cell (WBC) count (20.24×10^9/L), a decreased eosinophil count (0), and an evaluated C-reactive protein level (255.3 mg/dl). Other laboratory data were within the normal range. The chest computed tomography (CT) revealed encapsulated and loculated pleural effusion in the left lower lobe (Fig. 1a, b).

Thoracentesis revealed that the pleural effusion was yellowish in color, positive in Rivalta's reaction, and contained numerous inflammatory cells (multinucleated cells > mononuclear cells). The total protein (564.00 mg/L), lactate dehydrogenase (4794.00 IU/L), and adenosine deaminase (93 IU/L) in pleural effusion were elevated, while carcinoembryonic

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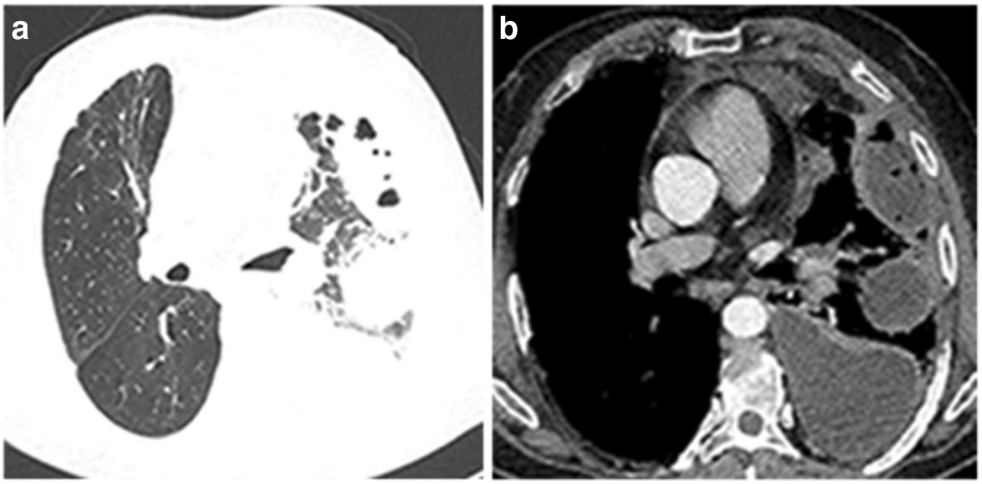

Fig. 1 Initial chest CT showed encapsulated and loculated pleural effusion (**a**, **b**)

antigen (1.89 μg/mL) was within the normal range. No malignant cells were detected on cytological examination of the pleural effusion. Several vermiform organisms were found in the drainage bag accidentally on the second day after percutaneous catheter drainage insertion (Fig. 2). Under the suspicion of parasite infection, the patient's serum and pleural effusion were examined by multiple-dot ELISA to detect specific antibody against various parasite antigens including *Paragonimus*, *Filaria*, *Toxoplasma*, *Taenia solium cysticercus*, *Schistosoma*, *Trichinella spiralis*, *Spirometra mansoni sparganum*, and *Angiostrongylus*, but no positive results were found. The organisms showed no body cavity, and their surface tegument was ridged at irregular intervals when viewed under the microscope. Pathological examination of these organisms performed by the Parasitology Department of Nanjing Medical University showed thick eosinophilic tegument and subtegumental calcospherules (i.e., calcareous corpuscles), in line with the characters of the metacestode form of *Spirometra mansoni* (Fig. 3).

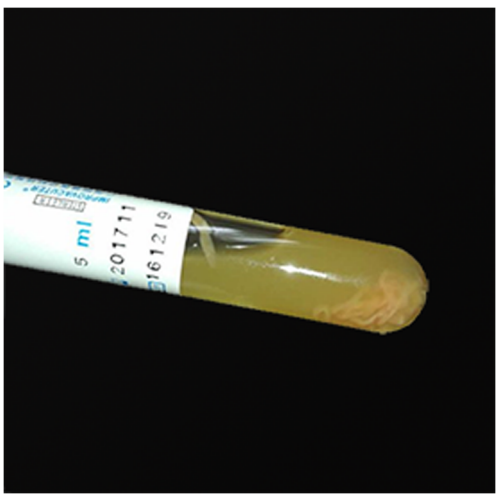

Fig. 2 Several vermiform organisms in the test tube that had the appearance of yarns

According to all these test results, this patient was diagnosed as having pleural sparganosis. He was treated with praziquantel at 75 mg/kg/day for 10 days, in combination with dexamethasone (0.5 mg bid) and moxifloxacin (0.4 g qd) to prevent immunoreaction and bacterial infection. One month after treatment, the pleural effusion had almost disappeared on the chest CT (Fig. 4a, b), and WBC count (6.05×10^9/L) and eosinophil percentage (4.5%) were back to normal. Besides, the patient's clinical symptoms were markedly improved.

Discussion

Sparganosis is a common zoonosis caused by the metacestode form of *Spirometra mansoni* and related diphyllobothriidean cestodes. Eggs of the worm are voided by the final host in the feces and hatch as coracidia in fresh water. Coracidia are eaten by the first intermediate host planktonic microcrustaceans and develop into the procercoid stage. When the infected crustaceans are eaten by the second intermediate hosts, such as frogs, snakes, birds, or freshwater fishes (Zeng et al. 2012), the procercoid completes metamorphosis into a plerocercoid, also known as a *sparganum* in the second intermediate hosts. When the second intermediate hosts are eaten by the definitive hosts such as dogs and cats, the *sparganum* develops into an adult tapeworm in the intestinal tract. Humans are accidently infected by drinking contaminated water or eating undercooked frogs, snakes, or tadpoles infected with the procercoids or spargana (Liu et al. 2015a, b; Cui et al. 2017).

Since the first sparganosis case was diagnosed in 1882, human sparganosis has been reported sporadically around the world, mainly in Asian countries (Lu et al. 2014). Clinical manifestations of human sparganosis are diverse. Among them, slowly growing and migratory subcutaneous nodules are the most common symptoms, so that this disease is mainly diagnosed and treated by dermatologists (Bracaglia et al. 2015). The thoracic cavity is a rare site for the

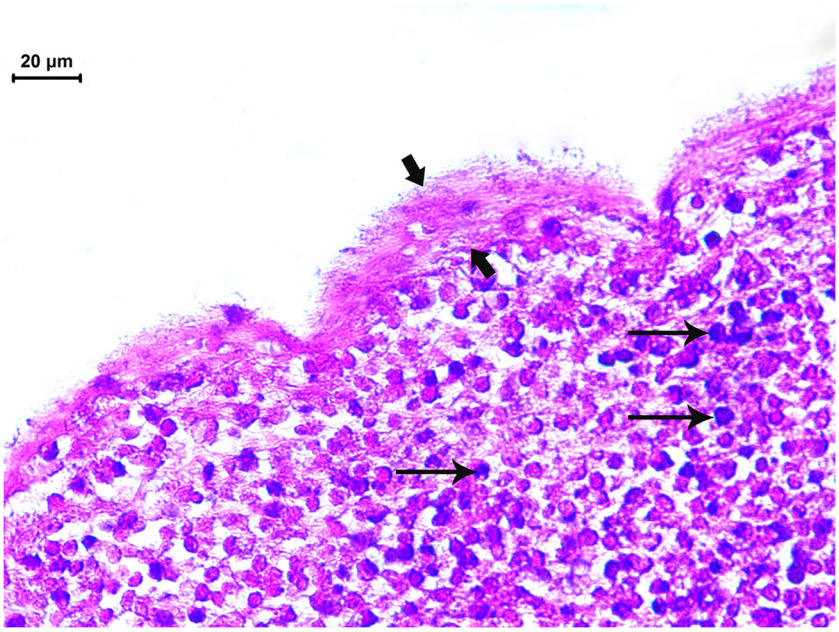

Fig. 3 *Sparganum* shows tegumental structure (short arrows) and calcospherules in the subtegumental layer (long arrows) (hematoxylin and eosin stain, Bar = 20 μm)

localization of this parasite in humans (Chen et al. 2015). The literature demonstrates that only 40 cases of pulmonary or pleural sparganosis have been described all over the world and are mainly distributed in China, Korea, Japan, and other Asian countries (Li et al. 2015). To our knowledge, about 10 pulmonary sparganosis cases have been reported in China, mainly distributed in Zhejiang, Guangzhou, Guangxi, and Yunnan provinces (Zhao and Wang 2014; Ouyang et al. 2014; Zeng et al. 2012). Our report here is the first case of pulmonary sparganosis occurring in Jiangsu province.

Although, he denied having eaten raw frogs, snakes, or any other undercooked flesh, as a manager of the farmers' market, he was exposed to live frogs or snakes in daily work. The chance of infection will increase with poor hand hygiene during the sampling inspection. The *sparganum* isolates from frogs collected in Jiangsu province have been identified as *sparganum* of *S. mansoni* (syn. *S. erinaceieuropaei*) using complete cytb and cox1 genes (Zhang et al. 2015). So, he was presumed to be infected through contact or accidental ingestion of the cestodes. Regarding the route of infection to the thoracic cavity, it is proposed that the parasite penetrated the intestinal wall and invaded the thoracic cavity through the diaphragm (Takeda et al. 2016).

Clinical information is not always helpful in making a definite diagnosis because the manifestations are nonspecific. Therefore, a serological or histological examination is important for the diagnosis of such complicated pulmonary infection (Cui et al. 2011a, b; Liu et al. 2015a, b). The presented patient was misdiagnosed as having bacterial pneumonia until the worms were found in the drainage bag and confirmed by histological examination. An ELISA test was also used for the diagnosis, but the result was negative. Accordingly, the

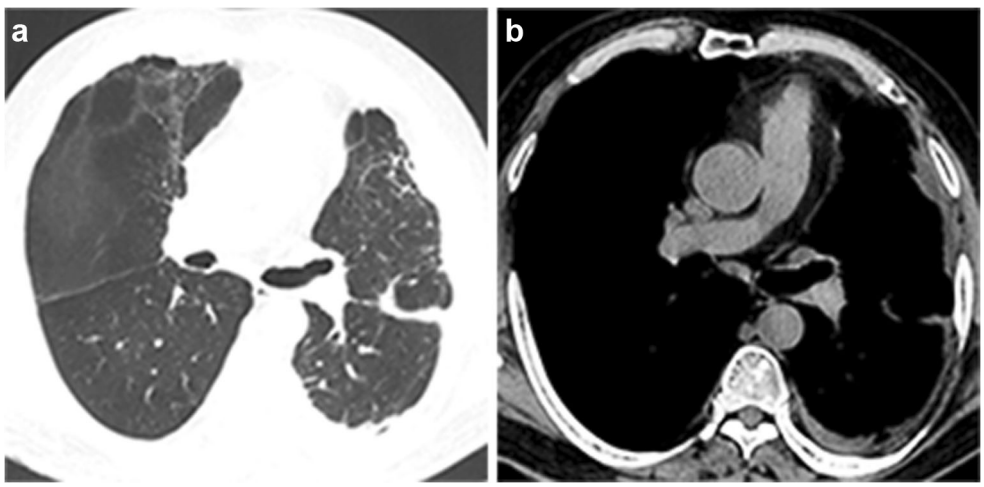

Fig. 4 Chest CT obtained 1 month after treatment showed pleural effusion significantly decreased (**a**, **b**)

patient has been suffering from asthma for years, and he has been treated with prednisone for a long period. Glucocorticoid has a general suppressive effect on immunity response, and antibody generation would be inhibited by extended glucocorticoid therapy (Daley and Peter 2015). The majority of patients with sparganosis show eosinophilia in peripheral blood and pleural effusion. But, the eosinophil count was decreased in peripheral blood in this case, which can be explained by extended glucocorticoid treatment.

The patient had no response to antibiotic therapy originally. After he was treated with oral praziquantel 75 mg/kg/day for 10 days, his clinical symptoms were relieved and the pleural effusion reduced gradually. Although sparganosis is usually related to raw food diet, the occupational exposure must be taken into consideration, especially for those working in a farmers' market or contacting live snakes or frogs in daily work. So, clinicians should acquire the history of disease entirely, especially the occupational exposure.

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Compliance with ethical standards

This study was approved by the Medical Ethics Committee of Nanjing Medical University (Nanjing, Jiangsu, China), and all the procedures involving human participants were performed in strict accordance with the 1964 Helsinki declaration and its later amendments.

Conflict of interest The authors declare that they have no conflict of interest.

Informed consent Informed consent was obtained from all individual participants included in the study.

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