Sparganosis Presenting as Pericardial Effusion and Lung Lesions

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Abstract

We herein report a rare form of sparganosis in a 29-year-old man presenting with pericardial effusion and lung lesions. The diagnosis was confirmed by the patient's history of eating inadequately cooked snake, significant elevated eosinophils in the peripheral blood and pericardial effusion, and marked positive reactions against *Sparganum mansoni* antigen in the serum. After two consecutive doses of praziquantel treatment, the patient's symptoms and laboratory and imaging findings were improved. Both specific antibody detection and follow-up of the patient's eosinophils, serum antibody, and imaging changes are important for sparganosis diagnosis, particularly in cases without a subcutaneous lump or mass.

Key words: sparganosis, pericardial effusion, lung lesions

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Introduction

Sparganosis, caused by the plerocercoid of the tapeworm, has been reported sporadically around the world, especially in several Asian countries, including South Korea, Japan, Thailand, and China (1-4). Infection in humans occurs by drinking water contaminated with proceroid-infected copepods, ingesting raw or inadequately cooked flesh of frogs or snakes infected with spargana, or by placing poultices of frog or snake flesh or skin on the eyes or open wounds and other lesions.

The most common clinical manifestation of sparganosis is a subcutaneous lump or mass in the abdominal wall, scrotum, lower extremity, or chest wall. *Sparganum mansoni* can also invade muscle, intestines, breast tissue, eyes, spinal cord, brain and, rarely, the thoracic cavity, such as the lung (5), peritoneopleural cavity (6-13), and pericardium (14).

We herein report a rare form of human sparganosis that presented as pericardiopleural effusion and lung lesions without a subcutaneous mass or lump.

Case Report

A 29-year-old man was admitted to our hospital for progressive shortness of breath for two weeks in the middle of December, 2011. Approximately 3 weeks before admission, the man had a fever of 38°C and was relieved after two days of intravenous treatment with antibiotics. However, the patient felt a shortness of breath initially during exercise which gradually led to the onset of dyspnea while in the supine position. The symptom could be abated by sitting up or while in the lateral position. The patient's past history was unremarkable except he reported eating inadequately cooked snake 6 weeks prior to admission.

The patient's physical findings were as follows: blood pressure, 120/88 mmHg; heart rate, 110/min; respiratory rate, 20/min; body temperature, 37.4°C; jugular vein distention; enlarged cardiac dullness; and distant cardiac sound.

The initial laboratory test results were as follows: an increased white blood cell (WBC) count, eosinophil count, erythrocyte sedimentation rate (ESR), C-reactive protein (CRP) level, and total serum IgE level (Table 1). The results

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Table 1. Laboratory Data of the Blood

Variables	On Admission	On Day 2 after Admission	On Day 4 after Admission	Reference Range
White blood cells ($\times 10^9/L$)	10.77	11.64	11.65	4-10
Eosinophil Count (%)	11	18.9	30	0.5-5
Eosinophils ($\times 10^9/L$)	1.18	2.20	3.50	0.02-0.5
C-reactive protein (mg/dL)	6.1	26.8	N/A	0-3
Total serum IgE (IU/mL)	258	406	N/A	< 200
Lactate dehydrogenase (U/L)	241	254	N/A	109-245

N/A: not available

Figure 1. Images obtained at the time of admission. (A) Echocardiogram demonstrated pericardial effusion with a maximal depth of 36 mm without hemodynamic significance. (B) Chest CT showed a large amount of pericardial effusion and a minimal amount of pleural effusion and atelectasis in the left inferior lung lobe. PE: pericardial effusion, LA: left atrium, LV: left ventricle, RV: right ventricle, AO: aortic root

Table 2. Laboratory Data of the Pericardial Effusion

Variables	On Admission	On Day 2 after Admission	Reference Range
Color	Bloody	N/A	N/A
Rivalta's reaction	Positive	N/A	N/A
Red blood cells (/mm ³)	25,416	N/A	N/A
White blood cells (/mm ³)	1,512	N/A	N/A
Lymphocytes (%)	38	N/A	N/A
Neutrophils (%)	51	N/A	N/A
Eosinophils (%)	11	N/A	N/A
Specific gravity	1.022	N/A	N/A
Lactate dehydrogenase (U/L)	1,150	1,322	N/A
Total protein (g/L)	47.06	46.16	N/A
Sodium (mmol/L)	141	141	N/A
Potassium (mmol/L)	3.7	3.9	N/A
Chlorine (mmol/L)	103	104	N/A
Glucose (mmol/L)	6.7	6.7	N/A
Adenosine deaminase (U/L)	48	51	<45

N/A: not available

of urine and stool routine tests were normal, as were liver and renal function, electrolyte, thyroid function, tumor markers (including AFP, CEA, PSA, CA 19-9, CA 15-3, CA 242, NSE, and cyfra 21-1), cardiac troponin T (cTnT), cre-

atinine kinase, MB isoenzyme (CK-MB), serum autoimmune antibodies [including rheumatoid factor, anti-nuclear antibody (ANA), ds-DNA, extractable nuclear antigen (ENA), anti-neutrophil cytoplasmic antibodies (ANCA), anti-mitochondrial antibody (AMA), anticardiolipin, c-ANCA, and p-ANCA], enterovirus RNA, and Coxsackie B virus IgM. The purified protein derivative (PPD) test was also negative.

Electrocardiography (ECG) showed sinus tachycardia with T wave flat in lead of II, III, AVF, and V1-6. Echocardiography demonstrated a pericardial effusion of maximal depth 36 mm without hemodynamic significance (Fig. 1A). Chest computed tomography (CT) also showed a large amount of pericardial effusion and a minimal amount of pleural effusion and atelectasis in the left inferior lung lobe (Fig. 1B).

The pericardial effusion was bloody in color, positive in Rivalta's reaction, and contained numerous red blood cells (25,416/mm³) and inflammatory cells (1,512/mm³, 38% lymphocytes, 51% neutrophils, and 11% eosinophils). Protein and lactate dehydrogenase levels in the pericardial fluid and serum were 47.06 g/L and 1,150 U/L and 73 g/L and 241 U/L, respectively. Other pericardial effusion results were as

Figure 2. Chest CT obtained after pericardial effusion drainage (-2,000mL) showed a medium amount of pericardial effusion, a minimal amount of pleural effusion, and multiple lesions in the left inferior lung lobe with high CT values.

follows: specific gravity, 1.022; glucose, 6.7 mmol/L; sodium, 141 mmol/L; chlorine, 103 mmol/L; potassium, 3.7 mmol/L; and adenosine deaminase (ADA), 48 U/L (Table 2). Malignant cells were not detected by cytological examination. Cultures for *Mycobacterium tuberculosis* or other bacteria were negative. Follow-up chest CT after pericardial effusion drainage (-2,000 mL) showed a medium amount of pericardial effusion, a minimal amount of pleural effusion, and multiple lesions in the left inferior lung lobe with high CT values (Fig. 2).

We suspected a parasitic infection based on the elevated eosinophils in the peripheral blood and pericardial effusion. However, neither worms nor eggs were found in the patient's stool or pericardial effusion. His serum was therefore examined by Enzyme-linked immunosorbent assay (ELISA) to detect specific antibody against various parasite antigens including *Clonorchis sinensis*, *Paragonimus westermani*, *Schistosoma*, *Echinococcus granulosus*, *Trichinella spiralis*, *Toxoplasma gondii*, *Angiostrongylus cantonensis*, *Filaria*, *Sparganum mansoni*, and *Cysticercus cellulosae*. The patient's serum was positive for *Sparganum mansoni* antibodies.

Anti-*Sparganum mansoni* IgG of the patient's serum were determined using a commercially available ELISA kit (Kangbaide Biological Technology, Shenzhen, China, #S.m 080910) as previously described (14, 15). Anti-*Sparganum*-specific IgG in the patient's serum was higher (OD=0.93±0.02) than the negative control level (OD=0.15±0.01). However, the ODs of the patient's serum IgG antibodies against the other parasites were similar to the negative controls.

Furthermore, fundus examination and head and abdomen CT showed no evidence of eye, brain, or abdomen involvement. As a result, the clinical diagnosis was *Sparganosis mansoni* infection. Because the left lower lung lesions were close to the diaphragm and ribs, it was hard to perform a transcutaneous lung biopsy or bronchoscopy to get a pathological diagnosis. We recommended a surgical resection of the lung, but the patient refused this suggestion.

The patient was therefore treated with praziquantel at 16.7 mg/kg/day for 9 days in combination with dexamethasone

(0.75 mg tid) and moxifloxacin (0.4 g/day) to prevent immunoreaction and bacterial infection. The patient was discharged after 9 days of treatment with a symptomatic improvement.

Approximately 10 days after discharge, the patient's blood examination showed a WBC count of $11.6 \times 10^9/L$ with 12.7% eosinophils ($1.47 \times 10^9/L$). The pericardial effusion decreased to less than 10 mm depth on echocardiography (Fig. 3A) and chest CT showed significantly decreased lung lesions (Fig. 3B).

Approximately 4 weeks later, the patient was treated with a second course of praziquantel (50 mg/kg/day) for three days with dexamethasone (0.75 mg tid). His eosinophil count gradually returned to the normal range (Fig. 4). Approximately 6 weeks after the second course of praziquantel treatment, both the pericardial effusion and the lung lesions disappeared on chest CT (Fig. 5). The IgG antibody against *Sparganum mansoni* remained positive, but the serum OD was much lower than before (OD of the serum: 0.45 ± 0.01 , OD of negative control: 0.13 ± 0.02).

After 2 years follow-up, the patient's eosinophil count, echocardiography, and chest CT all remained normal. Repeated ELISA showed the serum OD against *Sparganum mansoni* antigen was negative (OD of the serum: 0.21 ± 0.02 , OD of negative control: 0.16 ± 0.02).

Discussion

In the present case, the patient suffered from progressive shortness of breath due to a large amount of pericardial effusion. Common causes of pericardial effusion are idiopathic, infection (bacterial, viral, fungal, or parasitic organisms), trauma, hemorrhage, cancer, radiation therapy, kidney failure, and autoimmune disorders (hypothyroidism, inflammatory bowel disease, and rheumatoid arthritis). Parasitic infection presenting as pericardial effusion and lung lesions in this patient was confirmed by the history of eating inadequately cooked snake, significant elevated eosinophils in the peripheral blood and pericardial effusion, marked positive reaction against *Sparganum mansoni* antigen in the serum,

Figure 3. Images obtained 10 days after the first course of praziquantel treatment. (A) Echocardiogram showed a minimal amount of pericardial and pleural effusion. (B) Chest CT showed a minimal amount of pericardial and pleural effusion and lung lesions significantly decreased. PE: pericardial effusion, LA: left atrium, LV: left ventricle, RV: right ventricle, AO: aortic root

Figure 4. The eosinophil count gradually decreased to the normal range after two courses of praziquantel treatment.

and effective treatment with praziquantel.

Sparganosis is one of the common zoonoses in Asian countries. The first case of human sparganosis was reported

in 1882 in Xiamen of Fujian Province, China. Currently, a total of more than 1,000 cases of human sparganosis have been reported in 25 provinces in mainland China (4, 16-18).

Figure 5. Chest CT obtained a month and a half after the second course of praziquantel treatment showed the pericardial effusion and lung lesions had disappeared.

Human sparganosis manifests with various clinical symptoms which typically manifest as slowly growing and migratory subcutaneous nodules. The thoracic cavity is a rare site for the localization of this parasite in humans. To our knowledge, less than 20 cases of pleural sparganosis in Asia (7-13) and only 1 case of recurrent pericardial effusion and pleural effusion have been reported (14).

Regarding the route of infection to the thoracic cavity, it is proposed that the parasite penetrates across the intestinal wall and invades the thoracic cavity through the diaphragm. Although a diagnosis of human sparganosis is usually established based on pathologic examinations of such worms, in our case it was impossible to perform a pathologically diagnosis due to the location of the lesions and the patient's refusal to undergo surgery. Concerning the treatment, praziquantel has been reported to be effective for the treatment of sparganosis-related pleural effusion (7-9) and pericardial effusion (14), which was confirmed based on the effectiveness of the treatment of our patient in the present case.

Although sparganosis in the thoracic cavity is rather rare, this possibility must be considered in cases of significantly elevated eosinophil counts in both the peripheral blood and pericardial effusion. Specific antibody detection by ELISA is helpful for making a differential diagnosis. Moreover, follow-up of eosinophil counts, serum antibody levels, and imaging changes following chemotherapy is also important for diagnosis, especially in cases lacking of pathological examination. Concerning the treatment, we suggest that praziquantel is a good therapeutic option for pericardial effusion and lung lesions related to sparganosis.

The authors state that they have no Conflict of Interest (COI).

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