Official Case Reports Journal of the Asian Pacific Society of Respirology
Respirology Case Reports
OPEN ACCESS
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Two cases of paragonimiasis westermani diagnosed after eosinophilic pleural effusion-induced hydropneumothorax

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Keywords
Paragonimus westermani, hydropneumothorax, eosinophilic pleural effusion.

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Received: 8 December 2015; Revised: 8 February 2016; Accepted: 26 February 2015

***Respirology Case Reports*, 4 (2), 2016, e00154**

doi: 10.1002/rcr2.154

Abstract

Our hospital is located in the Tono region in the southeastern district of Gifu Prefecture in which there are forests and inhabitants who still hunt and eat game meat. Therefore, boar meat increases the risk of contracting paragonimiasis. We treated two patients who were infected by *Paragonimus westermani* after eating boar meat. They developed hydropneumothorax in association with eosinophilic pleural effusion. For patients who have pneumothorax with concomitant pleural effusion and eosinophilia in the pleural fluid analysis, it is necessary to take a detailed history, which includes flesh food consumption and travel to an endemic area, and to make a careful examination while taking into consideration parasitic infections such as paragonimiasis.

Introduction

Paragonimiasis is a relatively rare endemic disease in Japan; however, a certain number of patients have the disease, and 30–40 new patients are diagnosed in Japan each year [1]. Imaging findings of the chest vary extremely [2–4]. It is frequently difficult to diagnose the disease unless doctors strongly suspect that a patient is positive for it. We herein report two patients who developed hydropneumothorax with eosinophilic pleural effusion and were diagnosed as having *Paragonimus westermani* infection.

Case Report

Case 1

A 14-year-old Japanese child visited the pediatrics department of our hospital in April 2013 because of left chest pain. She was referred to our department because of left hydropneumothorax on a chest X-ray. Blood test findings were a white blood cell count (WBC) of 6200/μL with 6.0% eosinophils, total immunoglobulin E level of 612 IU/mL, and C-reactive protein (CRP) level of 0.03/mL. Pleural fluid findings were an orange and extremely cloudy appearance, WBC of 30200/μL with 80% eosinophils, lactate dehydrogenase (LDH) level of 865 IU/L, and adenosine deaminase (ADA) level of 19.4 U/L. The results of bacterial and acid-fast bacterial culture and cytology were negative. Imaging findings of the chest were left pneumothorax, hydrothorax, and pulmonary infiltration (Fig. 1).

Case 2

A 55-year-old Japanese woman with left chest pain and cough was referred to and visited our department in May 2015. Blood test findings were WBC of 8100/μL with 18.7% eosinophils, total immunoglobulin E level of 66 IU/mL, and CRP level of 0.46 mg/mL. Pleural fluid color was same as WBC of 30,200/μL with 98% eosinophils, LDH level of 1230 IU/L, and ADA level of 35.8 U/L. The results of the bacterial and acid-fast bacterial culture and cytology were negative. Imaging findings of the chest were left pneumothorax, hydrothorax, and pulmonary infiltration (Fig. 1).

Clinical Course

Both patients underwent chest drainage because of pneumothorax, and because they had an extremely high eosinophil count in the pleural fluid, a detailed history of food

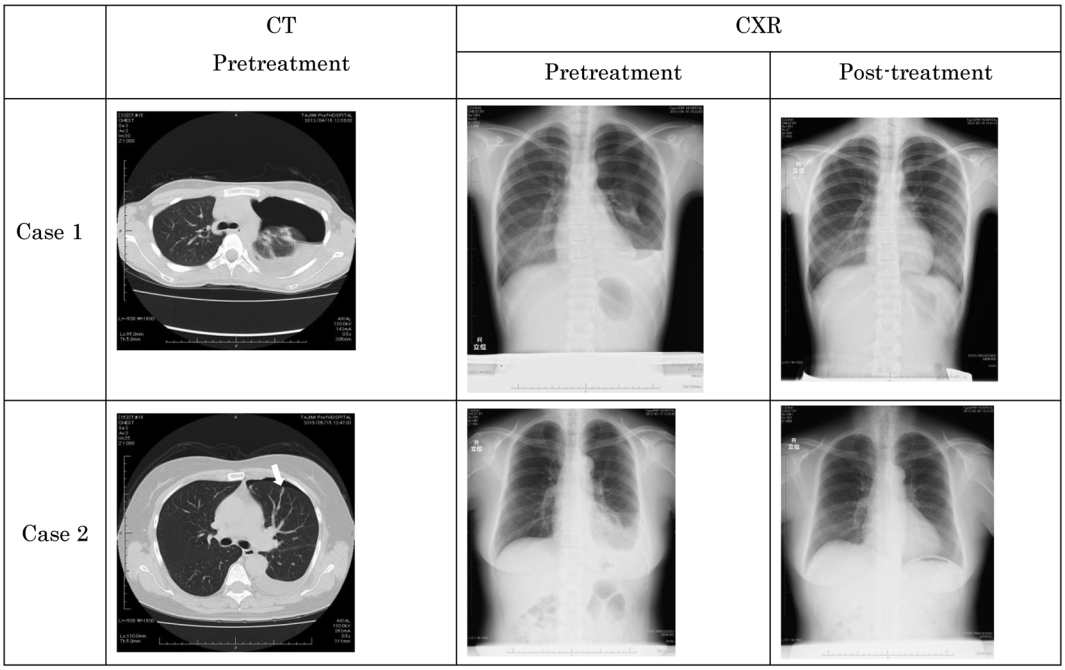

Figure 1. Chest computed tomography (CT) and chest X-ray (CXR) of pretreatment and post-treatment (from 3 to 4 months after) in case 1 and case 2. In case 2, CT detected a finding similar to worm migration tracts, probably crept in the lung (arrow).

consumption was obtained while taking into consideration paragonimiasis.

In case 1, the patient's family ate raw boar and venison. The patient cooked the boar meat that her husband hunted. The patients were assessed for the presence of antiparasitic immunoglobulin G antibody by using multiple-dot enzyme-linked immunosorbent assay (ELISA). The tests were confirmed as positive for *P. westermani*. An increased antibody titer, which was subsequently determined via microplate ELISA, established a definitive diagnosis of paragonimiasis westermani (Figure 2). After treatment with praziquantel administered at a dose of 75 mg/kg/day for 3 days without any side effects, their symptoms and imaging findings were consequently improved (Figure 1). The antibody titer decreased approximately 3–4 months later (Figure 2), and they have not shown any clinical symptoms since the treatment.

In case 1, zonal dermal findings with pain caused by *P. westermani,* which probably crept under the skin, were observed before administering the antiparasitic drugs. The skin color changed from red to yellow. No parasite eggs were detected in the sputum, feces, or pleural fluid in either patient.

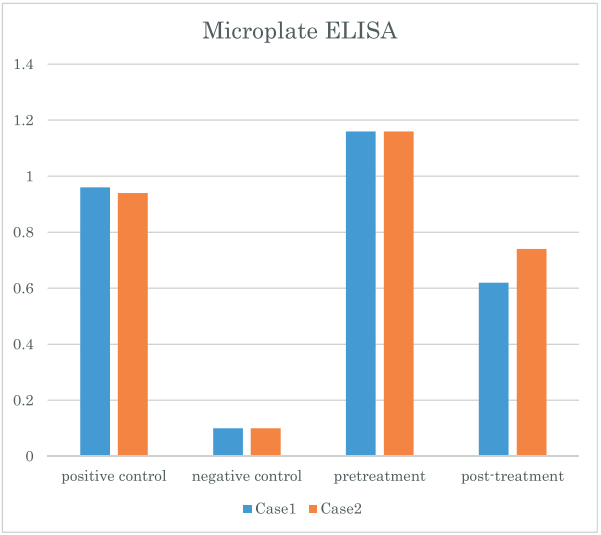

Figure 2. Antibody titer changes are shown. In both patients, the antibody titer at diagnosis is initially high and at 3 to 4 months after treatment decreases.

Discussion

Paragonimiasis westermani is a zoonotic parasitic disease caused by oral infection from heated or raw meat of intermediate hosts such as freshwater crab and Japanese mitten crab and from paratenic hosts such as the boar.

Nagayasu et al. [1] found that the respiratory symptoms of paragonimiasis westermani among patients were cough in 28.9%, sputum (including bloody sputum) in 27.3%, chest

pain in 18.5%, and respiratory distress in 10.4%. However, paragonimiasis frequently exists in asymptomatic patients who are confirmed as having an abnormality on routine chest radiography. Chest imaging findings of patients with paragonimiasis are diverse and range from pleural lesions (e.g., pleural thickening, pleural effusion, and pneumothorax) to intrapulmonary lesions (e.g., tubercles, invasive opacity, and cavitary lesions) [2–4]. Therefore, it is important to differentiate this disease from lung cancer or pulmonary tuberculosis.

A larva invading the lung causes pleuritis, pleural effusion, and pneumothorax in association with chest pain. Paragonimiasis is characterized by worm migration tracts, which indicate the trace of the parasite [2–4]. In computed tomography, a finding similar to worm tracts is detected.

Because hydropneumothorax occurs in diseases other than paragonimiasis, if we obtain the results of eosinophilic pleural effusion by routine pleural fluid tests, we may lead to diagnosis of the rare disease paragonimiasis. A definitive diagnosis of paragonimiasis is based on the presence of parasite eggs; however, the detection sensitivity of eggs in sputum, feces, and pleural fluid is low [2]. A serologic diagnosis by using microplate ELISA, as in this report, is useful and extremely reliable for paragonimiasis [5].

If paragonimiasis westermani is confirmed in a patient, the incidence of infection within a patient's family is high. Therefore, it is necessary to perform screening and antibody tests and to eliminate parasites as needed if a family member, although asymptomatic, has a history of eating infected food. In case 1, the patient's mother had hydrothorax 10 years before and was treated in a hospital. In case 2, the patient's asymptomatic daughter underwent tests at another hospital and was found to have pleural effusion associated with a high antibody titer. Worms are believed to die after heat cooking. However, insufficiently washed tools may cause infection, which occurred in case 2.

In conclusion, we experienced two cases of paragonimiasis westermani in which hydropneumothorax developed in association with eosinophilic pleural effusion. For a patient with hydropneumothorax, it is necessary to conduct a pleural fluid tests and if eosinophilic pleural effusion obtains in thoracentesis, to take a detailed history, which includes food consumption and travel to an endemic area, and to make a careful examination while taking into consideration parasitic infections such as paragonimiasis.

Acknowledgment

We are deeply grateful to Haruhiko Maruyama at the Division of Parasitology, Department of Infectious Diseases, and Faculty of Medicine at the University of Miyazaki (Miyazaki, Japan) for performing the immunological diagnoses.

Disclosure Statements

No conflict of interest declared.
Appropriate written informed consent was obtained for publication of this case report and accompanying images.

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