CASE REPORT

Active Hepatic Capsulitis Caused by Paragonimus westermani Infection

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Abstract

Paragonimiasis is an important re-emerging parasitosis in Japan. Although the lungs and pleural cavity are the principal sites affected with the parasite, ectopic infection can occur in unexpected sites such as skin and brain. This case report describes a patient with active hepatic capsulitis due to Paragonimus westermani infection. The patient was successfully treated with praziquantel at the dose of 75 mg/kg/day for 3 days.

Key words: paragonimiasis, lung fluke, ectopic infection, liver

Introduction

Paragonimiasis caused by infection with lung flukes, Paragonimus westermani or P. miyazakii is an important food-borne parasitic zoonosis and has been re-emerging in Japan (1, 2). The lungs and/or pleural cavity are the principal sites to be affected with the parasites. Because of the complexity of the migration route of Paragonimus spp. in the definitive hosts, ectopic infection can occur in unexpected sites. Although cutaneous or cerebral paragonimiasis are the classically known forms of ectopic paragonimiasis (3, 4), hepatic paragonimiasis is a rather rare form of ectopic infection. This case report describes a patient with active hepatic capsulitis caused by P. westermani infection. The patient was successfully treated with praziquantel (PZQ).

Case Report

This 44-year-old man, a public officer, was admitted to the Department of Radiology, Kagoshima City Medical Association Hospital for the purpose of lymph node biopsy in December 2000. The patient had a history of abdominal distension, right hypochondralgia and fever of approximately 2 months. He had been followed up for chronic hepatitis C for several years. Abdominal computed tomography (CT) revealed the presence of cholecystitis, hepatic capsulitis, liver abscess, and abdominal lymph node swelling (Figs. 1A and 1B). Lymph node swelling was also noted by chest CT in the left subclavicular and parasternal nodes. Peripheral blood leukocytosis (10,000/mm³) with moderate eosinophilia (14.5%) was noted by laboratory examinations. Total IgE level in serum was also elevated to 7,047 IU/ml. Laboratory data on admission revealed slight abnormalities in liver functions (Table 1), which were thought to be due to chronic hepatitis C. Histopathological examination on the biopsied lymph nodes revealed edematous inflammation without malignant cells or eosinophil infiltration. From these findings and the patient’s history of occasional consumption of undercooked wild boar meat, he was suspected of having a parasitic disease, most likely paragonimiasis or fascioliasis. Parasite eggs were not detected in stool. Cyst fluid aspirated from a subcapsular cystic lesion was serous, yellowish-white in color, and apparently purulent. Bacteriological or cytological analyses were not performed on the aspirated fluid. His serum as well as the purulent aspirate from the subcapsular abscess were positive against P. westermani antigen by multiple-dot ELISA (Fig. 2). He was given oral doses of PZQ 75 mg/kg/day for 3 days January 5–7, 2001. His symptoms rapidly eased and the eosinophil count was reduced to 6.2%. Hepatic capsulitis and subcapsular abscess disappeared 3 months after chemotherapy by abdominal CT observations (Figs. 1C and 1D). Efficacy of PZQ treatment was confirmed by the reduction of antibody titer in the sera before and after chemotherapy (Fig. 3).

Discussion

When patients with space-occupying lesions in the liver associated with eosinophilia and elevated total IgE level are encountered, the most probable disease to be considered is fascioliasis. In the present study, the patient is living in Kagoshima Prefecture, southern Kyushu, Japan, where both paragonimiasis (1, 2) and fascioliasis (5) are endemic. Thus, differential diagnosis between them was critically important and it was successfully made by immunoserological screening test using
Figure 1. CT scan on admission showed the presence of liver abscess (arrow in A), cholecystitis (arrow in B), and hepatic capsulitis (arrow in B) and abdominal lymph node swelling (arrowheads in B). The lesions had disappeared 3 months after PZQ treatment (C and D).

Table 1. Laboratory Data Related to Liver Function

<table>
<thead>
<tr>
<th>Test</th>
<th>Value</th>
<th>Abnormal Values</th>
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</thead>
<tbody>
<tr>
<td>Total bilirubin</td>
<td>0.9 mg/dl</td>
<td></td>
</tr>
<tr>
<td>Aspartate aminotransferase (AST)</td>
<td>44 IU/l</td>
<td></td>
</tr>
<tr>
<td>Alanine aminotransferase (ALT)</td>
<td>74 IU/l</td>
<td></td>
</tr>
<tr>
<td>γ-glutamyl transpeptidase (γ-GTP)</td>
<td>95 IU/l</td>
<td></td>
</tr>
<tr>
<td>Cholin esterase (Ch-E)</td>
<td>367 IU/l</td>
<td></td>
</tr>
<tr>
<td>Albumin</td>
<td>5.7 g/dl</td>
<td></td>
</tr>
</tbody>
</table>

Abnormal values are underlined.

Figure 2. Multiple-dot ELISA test of the patient’s serum and hepatic cyst fluid against parasite antigens. The patient’s serum and the cyst fluid were diluted to ×500 and ×200, respectively. NHS: normal human serum (positive control for the 2nd antibody), Di: Dirofilaria immitis, Tc: Toxocara canis, As: Ascaris suum, Asx: Anisakis simplex, Gd: Gnathostoma doloresi, Sr: Strongyloides ratti, Pw: Paragonimus westermani, Pm: Paragonimus miyazakii, Fh: Fasciola hepatica, Cs: Clonorchis sinensis, Se: Spirometra erinacei, Cc: Cysticercus cellulosae.
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Figure 3. Specific antibody titer in the sera before and after treatment.

multiple-dot ELISA (Fig. 2). Because PZQ is highly effective on paragonimiasis as shown in this study but not always effective on fascioliasis (6), immunodiagnosis must be done at an early stage of suspected parasite infections in the liver.

The migration route of Paragonimus spp. has been extensively studied and reviewed by Japanese parasitologists (3, 4). When metacercariae are ingested by humans or other definitive hosts, they excyst in the upper intestine and penetrate into the abdominal cavity. The juvenile worms migrate into abdominal muscles and lodge there for 5–7 days and come back into the abdominal cavity. Then they migrate through the diaphragm and the pleural cavity and finally reach the lung where they become mature adult worms. During the peritoneal stage, the juvenile worms often cause damage to the liver capsule and parenchyma. Yamagiwa (7) reported in his classical study that inflammatory lesions are frequently found in the hepatic capsule of the autopsied human paragonimiasis cases. The present patient had suffered from hepatic capsulitis for over 2 months without pulmonary involvement. Previously we reported a case of chronic occult intrahepatic paragonimiasis diagnosed by detection of P. westermani eggs in histopathological sections of intrahepatic cholelithiasis (8). Occult cases are also found accidentally as chronic granulomatous lesions in the peritoneal cavity (9). Based on the clinical study of P. skrjabini infections in humans and an experimental study of P. skrjabini and P. westermani infections in dogs, Hu et al (10) reported that the frequency of hepatic involvement in paragonimiasis is far higher than that expected. Thus, although humans are the definitive host for P. westermani, juvenile worms in the peritoneal cavity frequently migrate into the liver before they reach the lungs and sometimes fail further migration towards the lungs to cause the unusual disease in the liver.

In conclusion, the present results clearly show that the liver must be included in the possible affected site of P. westermani infection and that differential diagnosis between hepatic paragonimiasis and fascioliasis can be easily made by immunoserological tests.

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References