

## A case of subcutaneous paragonimiasis presented with pleural effusion

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### = Abstract =

Paragonimiasis is a parasitic infection that occurs following the ingestion of infectious *Paragonimus metacercariae* from crabs or crayfish. Pulmonary paragonimiasis is the most common clinical manifestation of this infection, but several ectopic paragonimiasis cases have also been reported. Among them, cases of subcutaneous paragonimiasis are rare, especially in children. We report a case of subcutaneous paragonimiasis of the right abdominal wall with pleural effusion with hepatic involvement and without abnormal pulmonary infiltration in a boy aged 2 years and 5 months. He had eaten soybean sauce-soaked freshwater crabs (kejang) 6 months prior to complaining of right abdominal wall distension. On evaluation, right pleural effusion without abnormal pulmonary infiltration was detected, as well as blood eosinophilia, an elevated serum IgE level, pleural fluid eosinophilia and a positive enzyme-linked immunosorbent assay that detected *P. westermani* antibody in the serum. Thoracentesis, praziquantel administration, and excision of subcutaneous lesions were performed. After treatment, the eosinophil count and serum IgE level were decreased, and the subcutaneous lesions did not recur. The frequency of paragonimiasis has decreased recently, but it is still prevalent in Korea. Paragonimiasis should be suspected if pleural fluid eosinophilia is associated with blood hypereosinophilia and a high level of serum IgE; however clinicians should obtain a thorough history of travel and food habits. (**Korean J Pediatr 2008;51:760-765**)

**Key Words :** Subcutaneous paragonimiasis, Pleural effusion, *Paragonimus westermani*

### Introduction

Paragonimiasis is a parasitic disease caused by *Paragonimus westermani* or other species of *Paragonimus* such as *P. miyazakii*, *P. uterobilateralis*, *P. mexicanus*, *P. ecuadoriensis*, *P. peruvianus*, *P. hueilungensis*, *P. skrjabini*, *P. heterotremus*, *P. philippinensis*, *P. kellicotti*, and *P. africanus*. Paragonimiasis is endemic in Korea, and three species, *P. westermani*, *P. pulmonalis*, and *P. iloktsuenensis*, are found in Korea<sup>1)</sup>. Until the late 1960s, paragonimiasis was prevalent in Korea, but the frequency of this disease has decreased recently because of ecological damage and changing attitudes. Cho et al.<sup>2)</sup> reported that the prevalence of human paragonimiasis in the 1990s was about 0.01 times the frequency

in the early 1970s in Korea.

The most commonly involved organ is the lung, but ectopic sites such as the brain, spinal cord, intestinal wall, peritoneal cavity, mesentery, liver, diaphragm, myocardium, and subcutaneous tissue can also be affected<sup>3)</sup>. Cutaneous paragonimiasis is rare, and only about 12 cases of cutaneous paragonimiasis without pulmonary infection have been reported<sup>4-14)</sup>. Moreover, no case of cutaneous paragonimiasis without pulmonary involvement in a child has been reported in Korea.

We report a case of subcutaneous paragonimiasis presenting as bulging masses of the abdominal wall with pleural effusion and hepatomegaly accompanied by serum eosinophilia in a child.

### Case report

A 2-year-5-month-old boy visited a local clinic due to bulging of the right abdominal wall. He was referred to

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Chonnam National University Hospital (CNUH) owing to the presence of hepatomegaly. The patient had no history of contact with tuberculosis. He and his family had eaten soybean sauce-soaked freshwater crabs (kejang) near the Seomjin River 6 months prior to visiting the clinic. He did not have any pulmonary system complaints such as a cough, sputum, dyspnea, chest pain, and hemoptysis or any abdominal symptoms such as abdominal pain and diarrhea.

On physical examination, he was found to have an approximately 3 cm sized palpable liver, palpable bulging mass on the right upper flank area and decreased lung sounds on the right side.

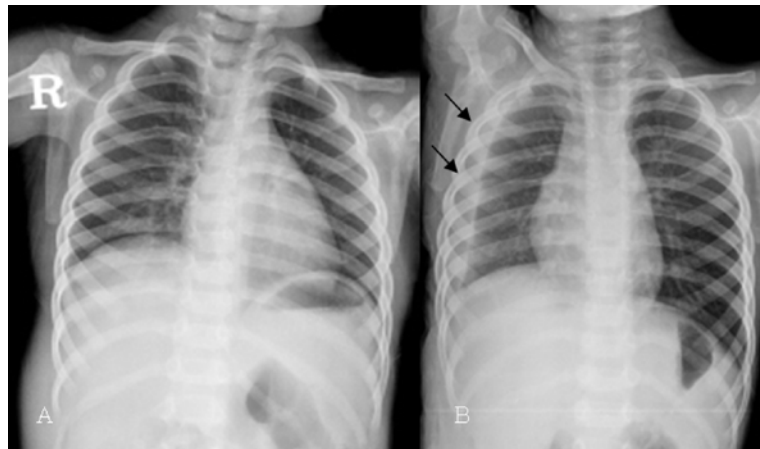
The results of laboratory studies were as follows: hematocrit, 38%; white blood cell (WBC) count, 28,100/mm<sup>3</sup> with 53.1% eosinophils and elevated serum IgE level, 825.0 IU/mL. The results of a complete chemistry panel to evaluate electrolytes and liver and kidney functions were within normal ranges. Muscle enzymes including creatine kinase (CK),

aspartate aminotransferase (AST), and alanine aminotransferase (ALT) were normal, but lactate dehydrogenase (LDH) was elevated to 864 U/L. The C-reactive protein (CRP) level was <0.2 mg/dL. An increase to 1:640 in a *Mycoplasma pneumoniae* antibody titer was detected.

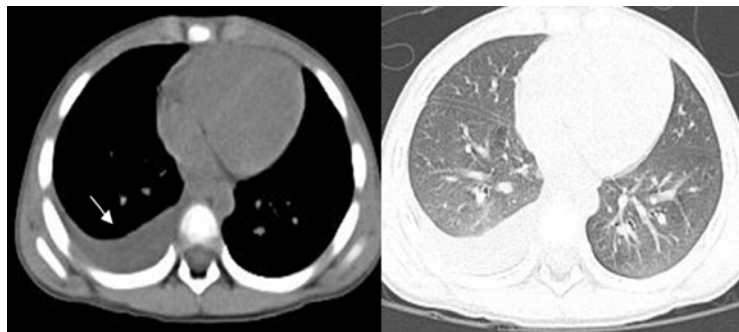
Sputum examination with Ziehl-Nielsen staining did not show bacilli. A purified protein derivative skin test for tuberculosis was negative.

### 1. Electrocardiography showed normal sinus rhythm

Chest roentgenogram and chest computed tomography (CT) on admission showed right pleural effusion, but abnormal findings in the lungs or lymph node swelling in the mediastinum were not observed. Abdominal ultrasonography detected a subtle increase in echogenicity of the liver with parenchymal liver disease with mild hepatomegaly was then suspected. Abdominal CT revealed multifocal, hypoattenuated lesions in the right hepatic lobe, a small subcapsular fluid



**Fig. 1.** Radiographs. A) Chest PA radiograph showing no abnormal pulmonary infiltration or mass shadow. B) Right chest decubitus radiograph showing pleural effusion (arrow).



**Fig. 2.** On the chest CT, abnormal pulmonary infiltration is not indicated, but right pleural effusion is seen (arrow).

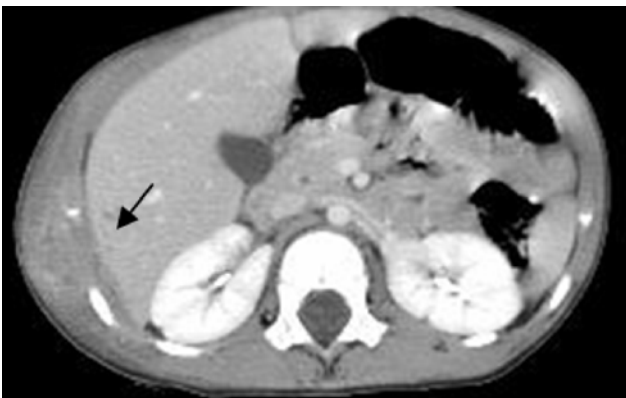
collection, and swollen right abdominal wall with edematous changes (Fig. 3).

Thoracentesis was performed for the right pleural effusion. The pleural effusion consisted of bright red-brown exudate with a pH of 7.322, protein concentration of 6.2 g/dL, high LDH of 1610 U/L, and WBC count of 27,500/mm<sup>3</sup> with 76% eosinophils. Parasites were not found.

The pleural effusion disappeared on chest roentgenogram, and the WBC count decreased to 15,300/mm<sup>3</sup> with 64.7% eosinophils.

An enzyme-linked immunosorbent assay (ELISA) for *P. westermanni* (GENEDIA<sup>®</sup> Cs/Pw Ab ELISA, Tecan Sunrise, Durham, NC) was positive. The patient was treated with praziquantel at 75 mg/kg for 3 days and was discharged.

One month later, he was seen in the outpatient department, and multiple subcutaneous nodules on the right flank were noted. The nodules had been gradually moving around. On physical examination, he was found to have right abdominal swelling, and the overlying skin in the right flank and inguinal area had risen slightly and was pigmented dark blue-brown (Fig. 4).



**Fig. 3.** On the abdominal CT, a swollen right abdominal wall with edematous changes is seen (arrow).

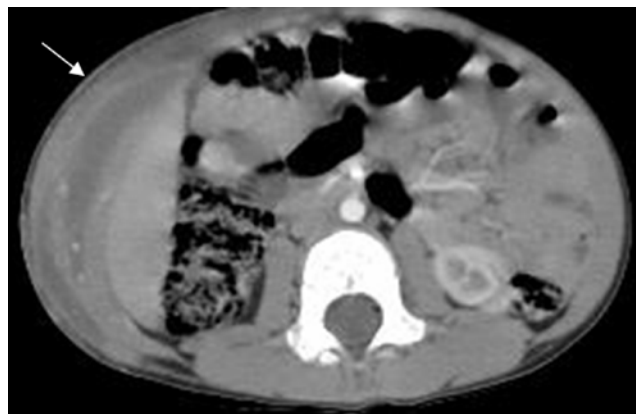


**Fig. 4.** Photograph showing right abdominal swelling. The covering skin has risen slightly and is pigmented dark blue-brown in the right flank and inguinal area (arrow).

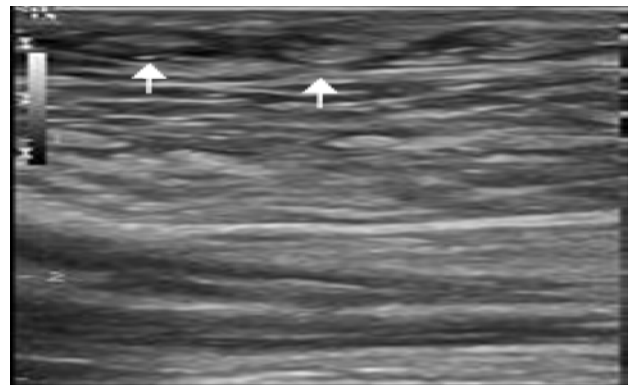
He was admitted again, and laboratory studies were performed. The hematocrit was 34.8%; the WBC count was 14,400/mm<sup>3</sup> with 28.4% eosinophils; the CRP level was normal at <0.2 mg/dL, and the IgE level was decreased to 175.0 IU/mL.

Abdominal CT showed more aggravation of the right abdominal wall lesions and a 15×2-cm intermuscular fluid collection and muscular swelling (Fig. 5). Abdominal ultrasound revealed multiple oval, peripheral hypoechoic lesions of the subcutaneous layer of the right abdominal wall, and a conglomerated area of edematous swelling and a focal inflammatory lesion were observed in the muscular layer of the right abdominal wall (Fig. 6). These findings were suggestive of paragonimiasis involving the abdominal wall.

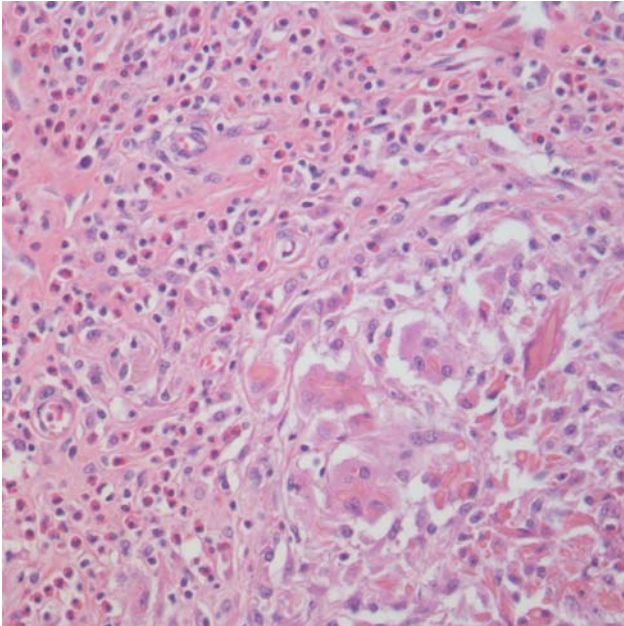
Excisional biopsy of the nodules was performed. The specimen showed inflammation with numerous eosinophilic



**Fig. 5.** On abdominal CT, a 15×2-cm intermuscular fluid collection and muscular swelling of the right abdominal wall are visible (arrow).



**Fig. 6.** The abdominal ultrasound shows multiple oval, peripheral hypoechoic lesions of the subcutaneous layer of the right abdominal wall and edematous swelling of the abdominal wall muscle layer (arrow).



**Fig. 7.** The biopsied specimen shows inflammation with predominant eosinophils (hematoxylin and eosin staining,  $\times 400$ ).

infiltrations, but parasites or other organisms were not found in the specimens (Fig. 7). The nodule was presumed to be a marker of migrating *P. westermani*. After treatment, the eosinophil count and serum IgE level decreased, and the cutaneous lesions, nodules, and swelling did not recur.

His family who eaten soybean sauce-soaked freshwater crabs (kejang) with him also performed enzyme-linked immunosorbent assay (ELISA) for *P. westermani*. The results were positive in grandmother and father. His father treated with praziquantel and thoracostomy due to pulmonary paragonimiasis with right pneumothorax and pleural effusion. His father also showed eosinophilia and elevated Ig E level in serum and pleural fluid. After treatment, he was cured.

## Discussion

More than 100 species of flukes, as either adults or larvae, infect humans. The most important of these in Korea is *Paragonimus westermani*, the lung fluke, which was discovered in the lungs of a Bengal tiger and classified by Kerbert in 1899<sup>15</sup>. *Paragonimus* infection is acquired by eating freshwater crabs or crayfish that are raw or inadequately cooked, salted, pickled, or soaked in wine or by eating cooked foods that are contaminated with viable larvae introduced from hands, utensils, or cutting boards used in the

preparation of crabs or crayfish<sup>3, 15</sup>. *Paragonimus westermani* is endemic in China, Taiwan, Korea, Japan, eastern India, Sri Lanka, Indonesia, and some areas of the former Soviet Union. Thus, carefully noting the patient history regarding travel and food habits is important for the diagnosis of paragonimiasis<sup>3</sup>. In our case, we did not initially check the dietary history. After a parasite infection was suspected, we asked about the dietary history, and his parents recalled that the family ate soybean sauce-soaked freshwater crabs (kejang) 6 months before his initial clinic visit.

The larvae of *Paragonimus* hatch from the eggs after at least 2 weeks of development in water. The free-swimming larvae penetrate a snail intermediate host and undergo development and multiplication for several weeks. Cercariae then emerge and encyst in the tissues of fresh-water crabs and crayfish. When humans or reservoir hosts ingest these second intermediate hosts, the larvae migrate across the small intestinal mucosa, enter the intra-abdominal space, and then penetrate the abdominal muscles. They return to the intra-abdominal space or migrate into the liver, and finally travel to the diaphragm and into the pleural space and lung. Approximately 2 to 3 months are required from the time of ingestion until the worms are fully mature. In most infections, the worms die within 10 years<sup>1, 3</sup>. Multiple lesions develop successively at different sites along the migration path of the larvae, and the development of multiple lesions suggests that a patient has ingested many metacercariae at once<sup>16</sup>.

*Paragonimus westermani* often causes an ectopic infection in various organs such as the brain, spinal cord, intestinal wall, peritoneal cavity, pleural cavity, mesentery, liver, adrenal gland, diaphragm, myocardium, and subcutaneous tissue<sup>1, 3, 17</sup>. Cutaneous paragonimiasis is very rare and is identifiable by the appearance of subcutaneous nodules, which may be fixed or migratory. Worms recovered from these lesions are immature<sup>3</sup>. The differential diagnoses are gnathostomiasis, sparganosis, and onchocerciasis<sup>7</sup>.

Several cases of cutaneous paragonimiasis with pleural effusion have been reported<sup>7, 11, 13, 18</sup>. In some of these cases, cutaneous symptoms occurred prior to pleural findings. In other cases, pleural effusion was followed by cutaneous paragonimiasis or both findings were detected simultaneously. In our case, pleural effusion was detected at the time of the patient's complaint of a bulging mass. If pleural fluid eosinophilia associated with blood hypereosinophilia is

detected, a cutaneous lesion should be detectable at that time or later.

Several different laboratory findings would suggest paragonimiasis. Eosinophilia is an important parameter in the suspicion of parasite infection at the early stages of infection, because it appears when the worms are alive and migrating. However, it disappears when the disease moves into its chronic phase, and thus a lack of eosinophilia does not necessarily rule out paragonimiasis<sup>19)</sup>. A definite diagnosis is made by the identification of eggs in sputum, feces, or pleural fluid or by the identification of the adult parasite or eggs in tissue. The identification of eggs in sputum, feces, or gastric juice is not easy in cases of mild, latent, therapeutically affected, or extrapulmonary paragonimiasis. Immunodiagnosis is useful in those cases, and an ELISA has been widely applied for the serologic diagnosis of paragonimiasis<sup>1, 3, 7, 15, 20)</sup>. In our case, a confirmed diagnosis was made by a positive ELISA result for *P. westermani*.

Ten kinds of drugs have been tried in experimental chemotherapy of paragonimiasis in Korea. Biothionol, niclofolan, and praziquantel are known to be effective for the treatment of paragonimiasis. Praziquantel at 75 mg/kg divided into three daily doses for 2 to 3 days has been regarded as the drug regimen of choice for paragonimiasis, resulting in a cure rate of 90 to 100%<sup>1, 3, 15)</sup>. Our case was treated with praziquantel.

Prevention involves education concerning the source of infection in endemic areas and changes in food habits. The frequency of paragonimiasis in Korea has decreased recently, but at least 10% of freshwater crabs sold in local markets are still infected with metacercariae of *P. westermani*<sup>2)</sup>. Consequently, efforts for the prevention of paragonimiasis need to be continued.

**한글 요약**

**홍수를 동반한 피하조직 이소기생 폐흡충증**

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폐흡충증은 갑각류나 생가재 내의 *Paragonimus*의 유충의 섭취로 발생하는 기생충 감염이다. 폐 폐흡충증이 가장 흔한 임상 양상이지만 몇몇 이소 폐흡충증이 보고되고 있다. 그 가운데 피하조직의 폐흡충증은 매우 드물다. 6개월 전 계장을 먹은 과거력이 있는 2세 5개월 남아가 우측 복벽의 종류를 주소로 내원하였

다. 흉부 X선과 CT 상 폐실질에 이상 없이 우측 흉수만 발견되었고 간비대를 보이고 있었다. 말초혈액 검사상 호산구 증가증을 동반한 백혈구 증가증 및 혈청 면역글로불린 E의 상승을 보였다. 이후 ELISA 상 폐흡충 항체에 양성반응을 보여 *P. westermani*에 의한 홍수를 동반한 피하조직의 폐흡충증으로 진단할 수 있었다. 환아는 praziquantel 복용으로 치료를 하였으나 피하조직의 폐흡충증 종류가 더 악화되어 이를 수술적으로 제거하였고 조직 검사상 기생충의 감염으로 의심되는 염증반응을 관찰할 수 있었다. 이에 본 저자들은 소아에서 감염된 피하조직의 폐흡충증의 드문 증례를 보고하는 바이다.

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