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Case Report

# *Paragonimus westermani* infection manifesting as a pulmonary cavity and adrenal gland mass: A case report☆

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ABSTRACT

We report a case of *Paragonimus westermani* infection simultaneously affecting two separate organs that presented as both a pulmonary cavity and adrenal mass in an immunocompromised host. A 65-year-old male with a previous kidney transplant visited our clinic because of hemoptysis. Chest computed tomography (CT) showed a pulmonary cavity and right adrenal gland mass. The *Aspergillus* antigen titer in bronchial lavage fluid was elevated and showed positive conversion. It was necessary to differentiate lung cancer with adrenal gland metastasis from a fungal infection with an adrenal gland adenoma. Positron emission tomography CT suggested benign disease, and it was misdiagnosed as pulmonary aspergillosis based on the elevated *Aspergillus* antigen titer in the bronchial lavage fluid. Owing to the adverse effects of anti-fungal treatment, the patient underwent wedge resection of the lung and *P. westermani* was confirmed. A careful history revealed that the patient had eaten raw freshwater crabs 3 years earlier, and a test for serum antibodies to *P. westermani* was positive. Despite treatment with praziquantel, the adrenal mass persisted on 3-month follow-up CT. A right adrenalectomy was performed and a *P. westermani* infection was confirmed.



## 1. Introduction

Paragonimiasis, or lung fluke disease is a parasitic disease of humans and other mammals that is caused by a trematode in the genus *Paragonimus* via the ingestion of raw, inadequately cooked crabs or crayfish infected with *Paragonimus* metacercariae. The parasite occurs in Southeast Asia, the Indian subcontinent, South and North America, and Africa. In Korea, freshwater crabs soaked in soybean sauce are the main source of human paragonimiasis. Paragonimiasis manifests in various ways including pleural effusions, pulmonary cavities, and pneumothorax [1–3]. The primary site of infection is the lung, although extrapulmonary involvement has been reported in the central nervous system (CNS), liver, peritoneal cavity, and abdominal wall [2,4]. Paragonimiasis has no specific symptoms or typical radiological findings, leading to the possibility of misdiagnosis [5]. In immunocompromised hosts, it can be misdiagnosed as a fungal infection or tuberculosis. It is also sometimes mistaken for a malignancy when it increases in size [6–8]. In areas where paragonimiasis is endemic such as Japan, India, and Korea, *Paragonimus* infection should be considered in the differential diagnosis of pulmonary cavitary disease and a careful history is necessary. We report a patient with a pulmonary cavity lesion and adrenal mass as a manifestation of *Paragonimus* infection. This is the first case of *Paragonimus* infection that simultaneously involved two separate organs, as confirmed by surgical resection.

## 2. Case report

A 65-year-old male presented with a history of blood-tinged sputum for several weeks. He had been treated with antitussives and oral antibiotics for presumed bronchitis, but the hemoptysis worsened. His vital signs were normal. Pulmonary auscultation detected wheezing in both lungs. The patient's white-blood cell count was normal, including the absolute number of eosinophils. His medical history included pulmonary tuberculosis with a complete course of anti-tuberculous medication 9 years earlier and

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renal transplantation 6 years earlier. Twenty months earlier, bilateral spontaneous pneumothorax was diagnosed and treated with oxygen supplementation and right chest tube insertion. At that time, there was no demonstrable cavitary lesion in either lung and no mass in the adrenal glands (Fig. 1A,B). Computed tomography (CT) of the chest when the patient presented with hemoptysis showed a pulmonary cavity with consolidation in the right lower lobe (RLL) and a 35-mm adrenal mass (Fig. 1C,D). Bronchoscopy was performed and empirical anti-tuberculous medication was prescribed because the patient had been taking immunosuppressive agents and had previously had tuberculosis. However, no organisms were seen on a Gram stain, acid-fast stain, or direct examination for fungal elements, and bacterial, mycobacterial, and fungal cultures were negative. Three months after starting the tuberculosis treatment, follow-up chest CT revealed no change in the pulmonary cavity with consolidation. The empirical tuberculous medication was stopped.

Nine months after presentation, the cavitary lesion in the RLL had grown slightly on CT (not shown) and was interpreted as an inflammatory lesion rather than a tumor. The right adrenal mass had not changed in size and intermittent coughing up with reddish material persisted.

One year after presentation, chest CT showed that the cavitary lesion had migrated from the superior segment to the lateral basal segment of the RLL and the right adrenal mass had grown to 40 mm (Fig. 1E,F). Positron emission tomography showed mild hypermetabolism (standardized uptake value $[SUV]_{max}$ 5.8) in the RLL lesion, which was considered to be a benign inflammatory lesion rather than a malignancy. The right adrenal mass showed intense hypermetabolism ($SUV_{max}$ 7.7), suggesting malignancy rather than a benign inflammatory lesion (Fig. 2). Hormone work-up for adrenal adenoma suggested a non-functioning tumor and a CT-guided adrenal mass biopsy was performed. This showed a dense eosinophilic infiltration with parasite eggs and no evidence of malignancy. A wedge lung resection was performed and the surgical specimen contained several ovoid structures and surrounding foreign body-type giant cells. The background showed lymphoeosinophilic infiltration and granulomatous inflammation (Fig. 3A).

After lung resection, we took a careful history and discovered that the patient had eaten raw crabs 3 years earlier. A serologic

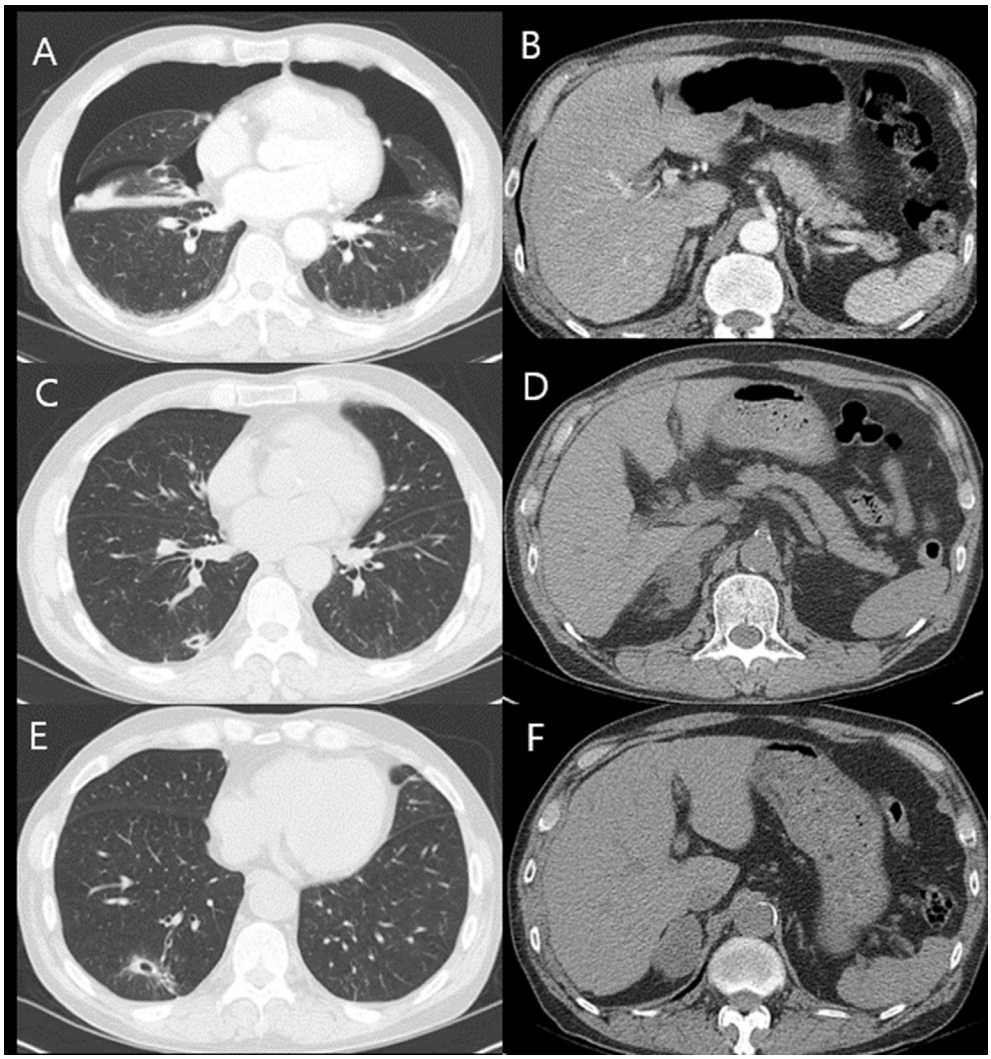


**Fig. 1.** Serial chest CT findings of pulmonary and intra-abdominal paragonimiasis in a 65-year-old male. Twenty months before the episode of hemoptysis, (A) CT shows a bilateral pneumothorax with no demonstrable cavity and (B) a scan of the upper abdomen showed no adrenal mass. On presentation with hemoptysis, CT shows (C) a subpleural cavitary nodule (arrow) in the RLL superior segment and (D) a newly developed 35-mm mass (arrow head) in the right adrenal gland. At the 1-year follow-up, CT shows (E) a slight increase in size and migration of the cavitary nodule (arrow) to the RLL lateral basal segment, and (F) the right adrenal mass (arrow head) had grown to 40 mm. CT, computed tomography; RLL, right lower lobe.

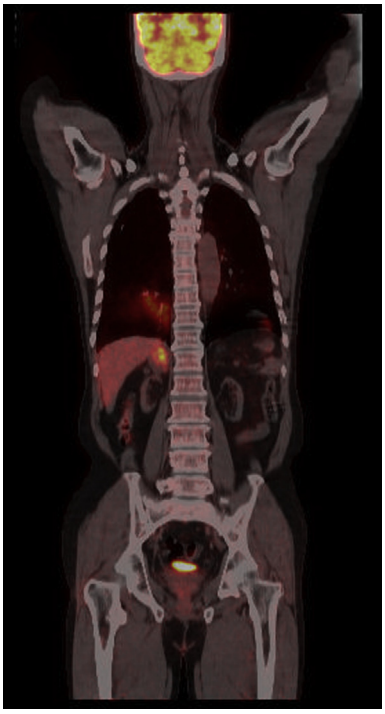

**Fig. 2.** A coronal positron emission tomography maximum intensity projection image showing diffuse hypermetabolic consolidation in the right lower lung (standardized uptake value $[SUV]_{max}$ 5.8) and a 44-mm hypermetabolic mass in the right adrenal gland ($SUV_{max}$ 7.7).

test for *Paragonimus westermani* (enzyme-linked immunosorbent assay; GC Labs. Yongin-si, South Korea) was positive. After 3 months of praziquantel treatment, follow-up abdominal CT revealed no change in the adrenal nodule. We decided to excise the right adrenal mass. A laparoscopic right adrenalectomy was done and the adrenal gland had an inflamed surface and was adherent to the adjacent tissues. Histologically, the adrenal lesion showed confluent geographic necrosis with a palisading granuloma and inflammatory infiltration. Remaining adrenal parenchyma could be seen (right-upper of image) (Fig. 3B). Several parasitic structures were present in the necrotic bed (Fig. 3C). These parasite eggs were elliptical and measured 85–110 μm, which is consistent with *Paragonimus* species (Fig. 3D).

## 3. Discussion

Here, we presented a case of concurrent pulmonary and intra-abdominal infection with *P. westermani* in a kidney transplant patient. There have been several reports of paragonimiasis in extrapulmonary sites [2,9] and multifocal lesions [4]. To our knowledge, however, this is the first report of concurrent *Paragonimus* infection in the lung and adrenal gland in which both sites were confirmed pathologically.

In our patient, the diagnosis was delayed because of the patient's underlying clinical condition. The consolidative cavity lung lesions were initially considered to be tuberculosis or a fungal infection because tuberculosis is endemic in Korea and the patient had been taking immunosuppressive drugs. Ultimately, paragonimiasis was confirmed surgically and serologically in the cavitary lung lesion and right adrenal mass.

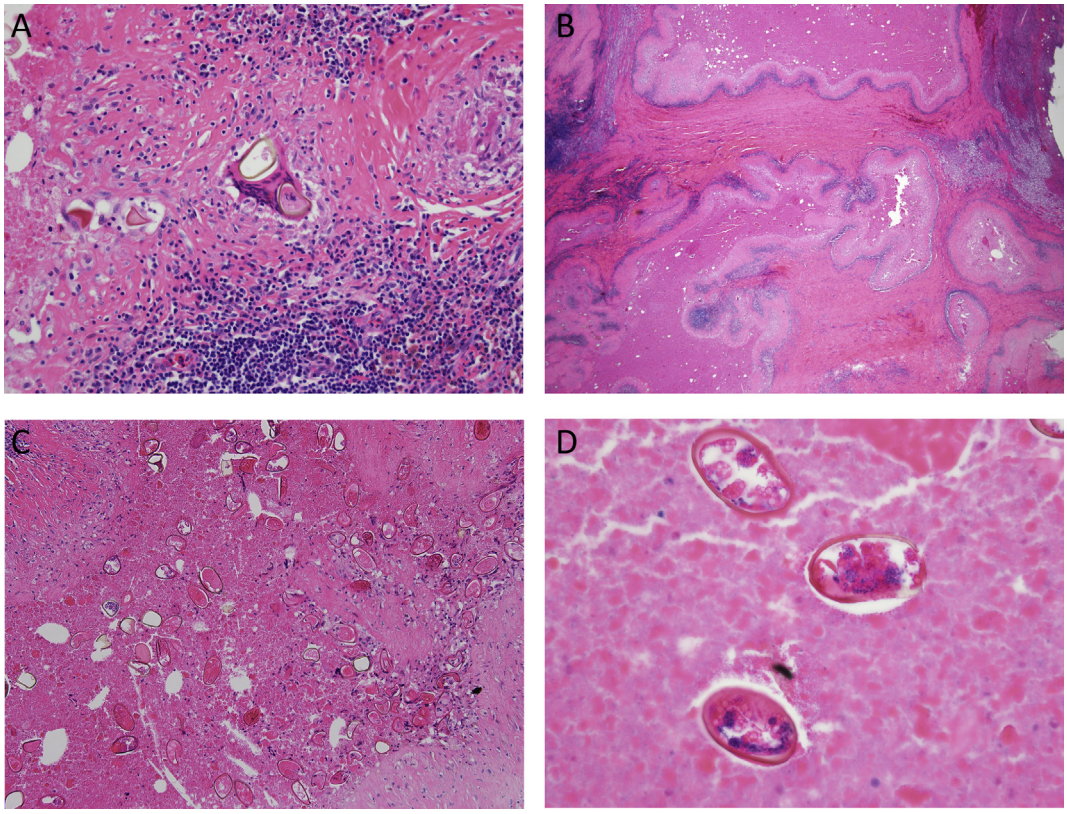


**Fig. 3.** Histological examination reveals (A) *Paragonimus* eggs in the lung parenchyma. (hematoxylin and eosin [HE] stain, x 200) (B) There is extensive necrosis of the adrenal parenchyma and sclerosis at a low magnification (HE stain, x 40) with (C) numerous parasite eggs in the necrotic bed (HE stain, x 100). (D) The eggs are ovoid and measure 85–110 μm (HE stain, x 400).

After ingestion by humans, the larvae of *P. westermani* penetrate the small intestine wall, enter the peritoneal cavity, and move through the diaphragm and pleural space into the lung. In the lungs, they form cyst-like capsules and mature inside the cavity and start producing eggs [10]. Metacercariae that migrate to the lung produce various clinical manifestations, including pneumonia, pleural effusions, and pneumothorax, as well as a variety of radiographic findings [11–13]. *Paragonimus* normally matures in the lungs of its hosts. However, atypical extrapulmonary migration of paragonimiasis is seen in the CNS, liver, peritoneal cavity and abdominal wall [2,4,14,15]. Although, the mechanisms of extrapulmonary paragonimiasis is not fully understood, there are a few hypotheses that explain the mechanism of extrapulmonary manifestations in *Paragonimus* infection. Calcified lung lesions and pleural thickening have been explained as associated with blocked passage of the metacercariae into the lungs [16]. Extrapulmonary manifestations can also develop while *Paragonimus* are seeking sexual partners, before establishment in the lungs [10]. It has also been hypothesized that when a small number of metacercariae are introduced into a host, they lack the power to invade the lung tissue and eventually manifest as an extrapulmonary infection [15]. The simultaneous development of multiple lesions has been explained as resulting from the ingestion of many metacercariae [4].

In areas where *Paragonimus* is endemic, paragonimiasis is usually diagnosed based on the patient's ingestion history, identification of eggs in the sputum, bronchial washings, gastric aspirate, or stool specimens, laboratory data, and immunodiagnostic tests [10]. In Korea, *P. westermani* is the only species causes human paragonimiasis and ELISA was positive for antibodies against *P. westermani* in our case [17]. In our patient, pathological confirmation was necessary because the increasing size of the lung and adrenal lesions suggested a malignancy. In this case, paragonimiasis was diagnosed by surgical resection of both sites and subsequent serological tests, in contrast with a previous case that showed only eosinophilic infiltration without eggs [4]. While intra-abdominal paragonimiasis has been reported [2,15,16], our case is the first report of paragonimiasis involving two separate sites, the lung and adrenal gland, simultaneously. Our patient also had a bilateral pneumothorax without an identified cause twenty months earlier. We suspect that the patient was already infected with *P. westermani* at the time of the pneumothorax.

In conclusion, although paragonimiasis rarely results in a consolidative cavitary lesion in the lungs and an adrenal mass, physicians must consider the possibility of paragonimiasis in endemic areas, and take a careful history of eating habits, while performing a histological examination and immunological tests.

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## Conflicts of interest

All authors claim no conflicts of interests.

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