

## Pleuropulmonary Paragonimiasis with Intrathoracic Mass

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**Pleuropulmonary paragonimiasis is a food-borne parasitic disease caused by the lung fluke *Paragonimus westermani* or other species of *Paragonimus*, which is endemic in Southeast Asia. It presents mainly pleural effusion or intrapulmonary nodules with respiratory symptoms. However, here we describe an exceedingly rare case of *Paragonimus westermani* with a mass in the pleural cavity. A 47-year-old man, who had presented with chest pain nine months earlier, was found to have right pleural effusion on detection survey computed tomography. He had a history of asbestos exposure and river fishing as a hobby and was confirmed to have *Paragonimus westermani* by immunodiagnosis. Because of a high level of hyaluronic acid in pleural effusion, he underwent a thoracoscopic examination. The pleura of the thoracic wall thickened greatly and showed no malignant lesion on biopsy. A white mass measuring 8 cm in diameter showed in the pleural cavity, which partially connected with the diaphragm and pulmonary pleura of the lower lobe. The postoperative pathological examination reported that the intrathoracic mass was a lesion that contained necrotic tissue enveloped with a fibrin capsule, which was thought to be formed by paragonimus. (Ann Thorac Cardiovasc Surg 2010; 16: 436–438)**

**Key words:** paragonimiasis, paragonimus, pleural effusion

### Introduction

Paragonimiasis is a foodborne parasitic disease common in Southeast Asia, especially in Japan, Korea, the Philippines, Taiwan, and parts of China.<sup>1)</sup> In Japan, two species, *Paragonimus westermani* and *Paragonimus miyazaki*, are known as the pathogens of human paragonimiasis, and most cases of paragonimiasis are infections resulting from *Paragonimus westermani*. It mainly presents as pleural effusion, intrapulmonary nodules, and pneumothorax with respiratory symptoms. However, here we describe a case

of *Paragonimus westermani* with a white mass measuring 8 cm in diameter in the pleural cavity, which partially connected with the diaphragm and pulmonary pleura of the right lower lobe.

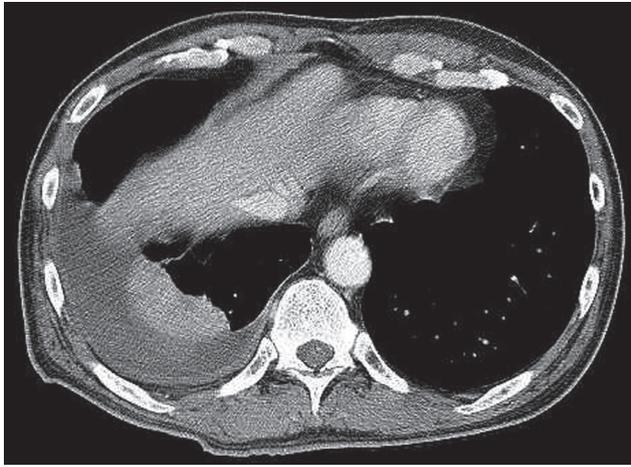
### Case Report

A 47-year-old man with chest pain was admitted to the Social Insurance Central General Hospital. Chest radiography showed right pleural effusion. He had a history of asbestos exposure and river fishing as a hobby. The heart-related disorder was negative on examination; thereafter he was simply followed. After two months, the pleural effusion decreased by conservative therapy. He demonstrated pleural effusion, and a mass in the right lower lobe on detection survey computed tomography repeated nine months after the first examination. Chest computed tomography showed a mass measuring 5 cm in diameter under the thickened pleura of the right lower lobe (Fig. 1). His laboratory examination on admission showed eosinophilic

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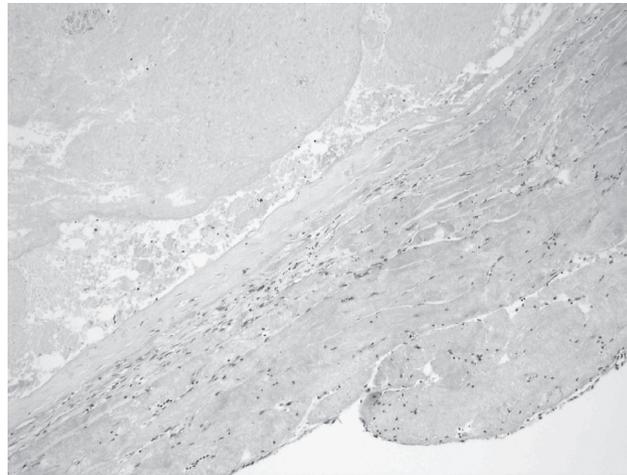
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**Fig. 1.** Chest computed tomographic scan on admission showing right pleural effusion and a mass under thickened pleura of the lower lobe.



**Fig. 2.** Thoracoscopic finding showing a white mass measuring 8 cm in diameter in the pleural cavity that partially connected with the diaphragm and pulmonary pleura of the right lower lobe.



**Fig. 3.** Photomicrograph of the mass in the pleural cavity showing necrotic tissue enveloped with a fibrin capsule without a body and without ova of paragonimus (PAS stained,  $\times 400$ ).

elevation of 6.4% of white blood cells. In pleural effusion, eosinophils were not elevated, but hyaluronic acid was 152,000 ng/ml. He was confirmed by immunodiagnosis to have *Paragonimus westermani*. He underwent thoracoscopic examination because of a high level of hyaluronic acid in pleural effusion. Thoracoscopic findings showed a white mass measuring 8 cm in diameter in the costophrenic sulcus, which partially connected with the diaphragm and pulmonary pleura of the right lower lobe (Fig. 2). The pleura of the thoracic wall thickened greatly and showed no malignant lesion on biopsy. Tumorectomy, partial resection of the right lower lobe, and biopsy of pleura were

performed. The pathological examination reported no malignant findings in the pleura, the mass, or pleural effusion. The intrathoracic mass was necrotic tissue enveloped with a fibrin capsule (Fig. 3). The body and ova of the paragonimus could not be found in the mass, pleural effusion, or pulmonary tissue. The pleural effusion culture showed no microbial detection. The mass density of the lower lobe on preoperative CT was diagnosed as atelectasis with high pleural thickness pathologically. The patient was diagnosed with *Paragonimus westermani* and treated with praziquantel for 3 days. He is being followed up under a warning of the onset of malignant mesothelioma.

## Discussion

The typical migration route of *Paragonimus westermani* in humans is as follows. When metacercariae, the infective stage of the parasite, are ingested by the final host, they excyst in the intestine and penetrate the abdominal cavity. The larvae migrate through the peritoneum, liver, diaphragm, and pleura into the lung, where they mature to adult flukes.<sup>1)</sup> The intrathoracic mass in this case connected closely with the diaphragm. Perhaps the larvae passed through this locus of the diaphragm. The larvae did not invade the pulmonary parenchyma from the pleural space and might subsequently have formed an intrathoracic mass and pus mass on the pulmonary surface. Cases of *Paragonimus westermani* showing a mass or pus mass in the pleural cavity are very rare. To our knowledge, only a few cases have been reported. Sawamura et al. reported a case of *Paragonimus westermani* with a tumor in the intrathoracic chest wall, which was yellow-white atheromatous material enveloped in a thick capsule.<sup>2)</sup> Kariatsumari et al. reported a case of *Paragonimus westermani* with fibrinopurulent material in the costophrenic sulcus and above the surface of the lower lobe.<sup>3)</sup>

There are comprehensive reports of the CT findings in *Paragonimus westermani*.<sup>4,5)</sup> According to these reports, pleuropulmonary paragonimiasis usually manifests as subpleural nodules that contain a necrotic low-attenuation area. In our case, there were pleural effusion and atelectasis with pus mass on vesicle pleura without intrapulmonary nodules. It is already known that only some of the larvae of *Paragonimus westermani* that arrive in the pleural cavity are parasitic on the lungs and grow to adult flukes. Fibrous histiocyte breeding soon occurs around adult flukes, and bag-shaped tissue called worm cysts is finally formed. Those formed in the pulmonary parenchyma present as nodules on radiographic examination, which at times have cavern-shaped findings. After adult flukes start laying ova in worm cysts, the ova of parasites can be detected in the expectoration of hosts. In our case, it is

possible that the larvae or adult flukes caused fibrous histiocyte breeding in the circumference of the pleural cavity, and a mass was formed. It is also possible that ova could not be detected because adult flukes could not form worm cysts and lay ova in the lung. For this reason, we might need to depend on serological diagnosis by a dot-ELISA method to detect parasite-specific IgG antibodies. Mukae et al. suggested that the dot-ELISA method for the detection of IgG antibodies is very useful for the diagnosis of paragonimiasis.<sup>6)</sup>

In our case, hyaluronic acid in pleural effusion was 152,000 ng/ml. Welker et al. reported that 100,000 ng/ml hyaluronic acid in pleural effusion correlated significantly with malignant mesothelioma.<sup>7)</sup> Our patient had a history of asbestos exposure; therefore he must be carefully followed up in the event that he should have an attack of mesothelioma in the future.

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