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Case Report

Two cases of pulmonary and pleural sparganosis confirmed by tissue biopsy and immunoserology

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ABSTRACT

Sparganosis in humans is an incidental infection and is known to be associated with eating insufficiently cooked meat of frogs and snakes or drinking unboiled stream water. Although it can involve various internal organs, pulmonary and pleural involvement due to sparganum is rare. Because we recently experienced two cases involving lung parenchyma and pleura that were misdiagnosed as bacterial pneumonia and lung cancer, we herein intend to present them in detail.

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Introduction

Sparganosis is a parasitic infection caused by the plerocercoid larvae of the genus *Spirometra*. It has a worldwide distribution, but the majority of cases have been reported in Japan, Korea, China and Southeast Asia.^{1,2} Humans are accidental hosts in the life cycle, while dogs, cats and other mammals are definitive hosts. Human infection mainly occurs by eating raw or insufficiently cooked frogs and snakes (second intermediate hosts), or by placing frog or snake flesh on open wounds for treatment of skin ulcers as well as on the eyes to treat inflammation.³ Sparganosis in humans usually appears as subcutaneous nodules all over the body and can involve the

eye, brain, and spinal cord.^{4,6} However, the involvement of lung and pleura is very rare. We herein present two cases of pulmonary sparganosis accompanied by pleural effusion and pulmonary nodules, which were confirmed by both histological and immunoserological examinations.

Case presentation

Case 1

In January 2010, a 57-year-old man was consulted at the department of gastroenterology due to an abnormality on a chest computed tomography (CT) that was performed

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seven days prior. He had chronic alcoholic pancreatitis and complained of left chest pain that had lasted for two weeks. He presented with fever, cough and sputum. The chest CT showed an ill-defined 4 x 3 cm mass-like consolidation with ground glass opacities, and an internal low attenuated lesion in the left lower lobe (Figs. 1A and 1B).

Under the suspicion of bacterial pneumonia, empirical antimicrobial therapy was started. Despite four days of therapy, his symptoms did not improve. He was transferred to the department of pulmonology. A chest x-ray obtained at the day of transfer revealed both small amounts of pleural effusion and increased opacity in the right middle lung zone. The left consolidation shown in the previous chest CT disappeared. Laboratory data showed 11,600/mm³ white blood cell count with increased eosinophil count (15%) and slightly increased C-reactive protein level (2.37 mg/dL). The result of pleural fluid analysis was an exudative effusion that was eosinophil-dominant (71%).

Another chest CT, performed seven days later, revealed decreased extent of the tract-like consolidation and residual nodular lesion with internal low attenuation in left lower lobe. Other newly developed tract-like consolidations in the right lower lobe were present, as well as pleural effusion and thickening (Figs. 1C and 1D). Under the suspicion of parasite migration, serum ELISA was performed to detect a parasite



Fig. 1 - Initial low-dose chest CT obtained at the time of consultation at the department of pulmonology (A,B). Prone position chest CT taken seven days after transfer (C, D). This demonstrated decreased extent of the tract-like consolidation and a residual nodular lesion with internal low attenuation in the left lower lobe, and other newly developed tract-like consolidations in the right lower lobe in addition to newly developed pleural effusion with pleural thickening, as compared with a previous CT.

infection. The patient's history was taken again, and it was noted that he had eaten wine-soaked snakes and had drunk unboiled stream water for the past three years. It was also revealed that he had eaten the flesh of a wild boar. Serum ELISA was strongly positive for sparganum.

To confirm the diagnosis and treatment, a video-assisted thoracic surgery (VATS) biopsy was performed in the right lower lobe. The larva was identified and removed. Histological findings demonstrated a sparganum with thick eosinophilic tegumentum and subtengumental calcospherules (Fig. 2A). The lung parenchyma showed eosinophilic parasite granuloma formation with a palisading histiocyte lining as well as central fibrinoinflammatory necrotic exudates. Scattered Charcot-Leyden crystals were also found (Fig. 2B). He was finally diagnosed with pulmonary sparganosis with pleural effusion. After surgical removal, he was given praziquantel. On the fifth day after removal, his serum eosinophil count was normalized and a follow-up chest X-ray showed no pleural effusion or pulmonary infiltration. A chest X-ray taken nine months after discharge showed no evidence of recurrence.

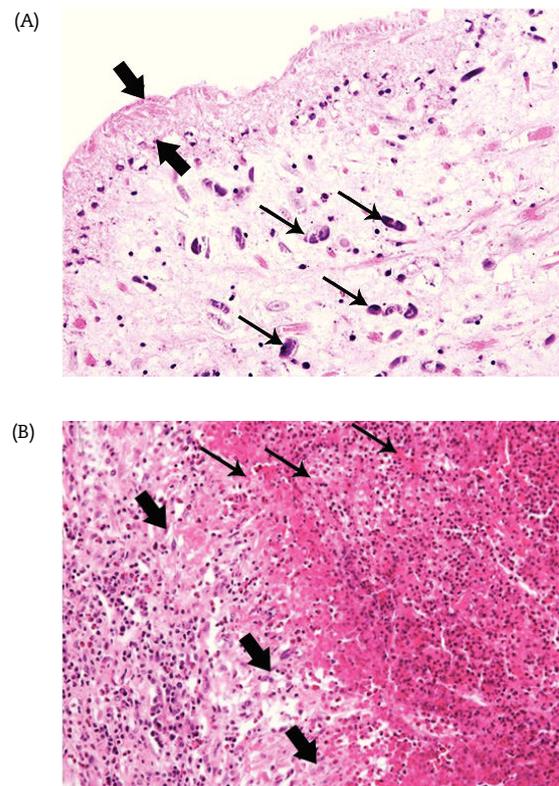


Fig. 2 - Sparganum shows a thick eosinophilic uniform tegumental structure (short arrows) and calcospherules (long arrows) in the subtengumental layer (A) (H-E stain, X 400). Parasite granuloma of the lung parenchyma shows palisading histiocytes (short arrows) lining the inner side with fibrinoinflammatory necrotic exudates and Charcot-Leyden crystals (long arrows). No worm is seen (B). (H-E stain, X 200).

Case 2

The patient was a 61-year-old man. He was referred to the department of pulmonology because of an abnormal pulmonary shadow on a chest X-ray in May 2011. He was a smoker (40 packs/year), and had no symptoms. There were no lesions anywhere on the body. A chest X-ray showed a small nodular opacity in the right upper lung. A chest CT showed a roughly 1.3 cm-sized, well-defined nodule with surrounding ground glass appearance in addition to a 0.6 cm-sized, well-defined nodule with interlobar fissural thickening in the right upper lobe and lower lobe, respectively (Figs. 3A and 3B). Laboratory data showed a $9,000/\text{mm}^3$ white blood cell count with slightly increased eosinophil count (6.0%) and increased C-reactive protein (4.37 mg/dL).

In order to rule out lung cancer, we performed a VATS biopsy in the right upper/lower lobes and pleura. A sparganum larva was seen migrating along the parietal pleura (Fig. 4). Histological findings of the parietal pleura and lung parenchyma demonstrated eosinophilic parasite

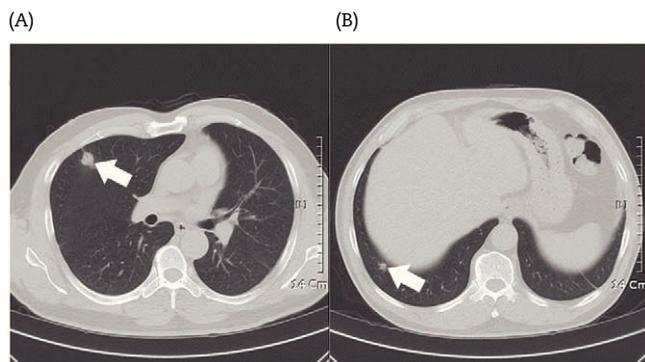


Fig. 3 - Initial chest CT demonstrated well-defined pulmonary nodules located in the right upper and lower lobes (white arrows).



Fig. 4 - Sparganum larva migrating along the parietal pleura found by thoracoscopy.

granuloma formation with a palisading histiocyte lining and central fibrinoinflammatory necrotic exudate. No worm was seen. The serum ELISA taken seven days after surgery was positive for anti-sparganum antibody. Intrapulmonary sparganosis was confirmed based upon these results. The patient's history revealed that he had drunk unboiled stream water.

Discussion

Sparganosis is an infection caused by the plerocercoid larvae (spargana) of various diphyllbothroid tapeworms belonging to the genus *Spirometra*. The larvae live in frogs and snakes, which serve as secondary intermediate hosts. Humans usually become infected by drinking contaminated water or ingesting undercooked frogs or snakes infected with the larvae.^{5,7} Although sparganosis has been reported sporadically all over the world, most of the cases occur in China, Japan, Korea and Thailand.⁸ This is associated with unusual habits of eating undercooked frog and snake flesh or drinking stream water in these countries. In humans, the larva invades the brain, spinal cord, breast and subcutaneous tissues. Recently, a human ocular sparganosis was reported in southern Brazil.⁹ It usually manifests as a migrating subcutaneous nodule and can result in blindness, paralysis, and even death. However, cases where there is invasion in the lung or pleura are rare. To the best of our knowledge, only six cases of pulmonary or pleural sparganosis have been described in the literature around the world.¹⁰⁻¹⁵ A Korean seroepidemiologic survey in a province with relatively higher prevalence rate showed a 3.3% positive rate for anti-sparganum antibody (IgG) in 719 adults, including two patients.¹⁶ Nonetheless, only one case of pulmonary sparganosis was reported in Korean literature.¹⁰

Recently, we experienced two cases of sparganosis invading both lung and pleura in our hospital. At first, the presented cases were misdiagnosed as bacterial pneumonia and lung cancer, respectively. The very low incidence rates of pulmonary or pleural sparganosis led to misdiagnose as other diseases. Detailed history of eating and drinking habits was not taken at the time of presentation, although both patients had a habit or history of eating uncooked snakes and/or drinking stream water. To confirm the diagnosis, both patients underwent surgery. The larvae migrating along parietal pleura were found by thoracoscopy. In addition, biopsy showed findings consistent with parasite granuloma including the worm. As a result, sparganosis was confirmed by thoracoscopic and histological findings as well as immunological results.

Anti-parasitic drugs such as praziquantel or mebendazole may be effective for sparganosis. However, complete extraction of larvae is a treatment of choice. The patient in the first case was followed for 10 months with two-month intervals. Clinical and radiological abnormalities were not detected during follow-up.

In differential diagnosis for pulmonary nodules or mass-like lesions on chest CT, pulmonary and pleural sparganosis should be considered, especially in regions with a high prevalence of food-borne parasitic zoonosis.

Conflict of interest

All authors declare to have no conflict of interest.

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