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Case Report

First case report of isolated pleural cysticercosis demonstrating pleuroscopic findings

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ABSTRACT

Pulmonary cysticercosis is a rare manifestation of human cysticercosis, which mostly occurs in developing countries. The disease can affect the lung parenchyma and pleura, resulting in pulmonary nodules, pneumonitis, lung cavities, or pleural effusion. We herein present a case involving a man of advanced age who presented with symptomatic eosinophilic pleural effusion. A computed tomography scan of the chest showed left pleural effusion without lung parenchymal abnormalities. Pleuroscopy revealed novel findings of cysticercal pleural nodules confirmed by histological examination. The patient was diagnosed with isolated pleural cysticercosis. His symptoms and chest radiographic abnormalities improved after treatment with anthelmintic drugs.

1. Introduction

Cysticercosis is an infectious disease caused by the parasite *Taenia solium* (pork tapeworm). When a human instead of a pig serves as an intermediate host, cysticercosis develops after ingestion of food or water containing parasite eggs. After penetration of the host's intestinal wall, the embryos enter the bloodstream to form larvae (cysticerci) in other body tissues, including the brain, eye, skeletal muscle, and subcutaneous tissue.

Pulmonary cysticercosis has been described in a few cases as a manifestation of disseminated disease. Patients may have respiratory symptoms including cough, shortness of breath, or pleuritic chest pain along with systemic symptoms such as fever and weight loss. However, isolated pulmonary cysticercosis is exceedingly rare. It is characterized by pulmonary nodules/infiltrates, lung cavities, and pleural effusion and has been reported in fewer than 10 cases worldwide [1–4]. We herein present a case of isolated pleural cysticercosis in which lung parenchymal involvement was absent but pleuroscopy revealed cysticercus-induced pleural nodules (see Figs. 1 and 2).

2. Case report

An 81-year-old man from Northeast Thailand presented with the chief concern of shortness of breath for 1 month. He also had a nonproductive cough, left-sided pleuritic chest pain, and a poor appetite. He denied fever or hemoptysis. His underlying disease was

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hypertension. He had no history of tuberculosis or high-risk contact. He did not smoke, and had no family history of lung cancer.

On examination, the patient was afebrile with respiratory rate of 20 breaths/min, and oxygen saturation was 96 % on ambient air. Examination of the respiratory system revealed dullness on percussion, decreased breath sounds, and positive egophony on the left side, corresponding to left pleural effusion occupying half the hemithorax. The remainder of the physical examination was unremarkable. Routine hematological and biochemical studies were normal apart from mild anemia (hemoglobin, 10.9 g/dL). Ultrasound-guided thoracentesis and closed pleural biopsy using an Abrams needle were performed. The pleural effusion was bloody in appearance and had a red blood cell count of 930,000/μL and corrected white blood cell count of 2730/μL (lymphocytes, 51 %; eosinophils, 25 %; neutrophils, 24 %). The adenosine deaminase level was 24.7 U/L. Cytologic examination of the pleural fluid and needle biopsy of the parietal pleura failed to establish a diagnosis (see Table 1).

Medical thoracoscopy under local anesthesia was performed for a definitive diagnosis. Pleuroscopy revealed diffuse pleural thickening with several small to large purplish pleural nodules (Fig. 2). Histological examination of the nodules demonstrated a fibrotic cyst wall with a degenerated scolex, consistent with cysticercosis (Fig. 3). The patient's dietary habit of ingesting raw pork and uncooked vegetables was revealed retrospectively. Eye examination and magnetic resonance imaging of the brain and eyes revealed no evidence of cysticercosis. The patient was treated with albendazole (15 mg/kg/day) and praziquantel (50 mg/kg/day) for 2 weeks, and his symptoms gradually improved. Another chest radiograph after 4 weeks of treatment showed significant radiological resolution (Fig. 4). The patient was additionally followed up at 3 and 8 months after therapy with no evidence of recurrence.

3. Discussion

Human cysticercosis occurs when pork tapeworm larvae grow within the human body. This disease is becoming a serious public health concern in many developing countries, especially Latin America, sub-Saharan Africa, and Asia, which are characterized by poor hygienic conditions. The World Health Organization has classified taeniasis/cysticercosis as a neglected tropical disease causing disastrous health and socioeconomic outcomes. More than 50 million people are infected worldwide, and approximately 50,000 deaths occur each year [5,6].

Neurocysticercosis, the most common type of cysticercosis, is correlated with serious morbidity and mortality. It affects the central

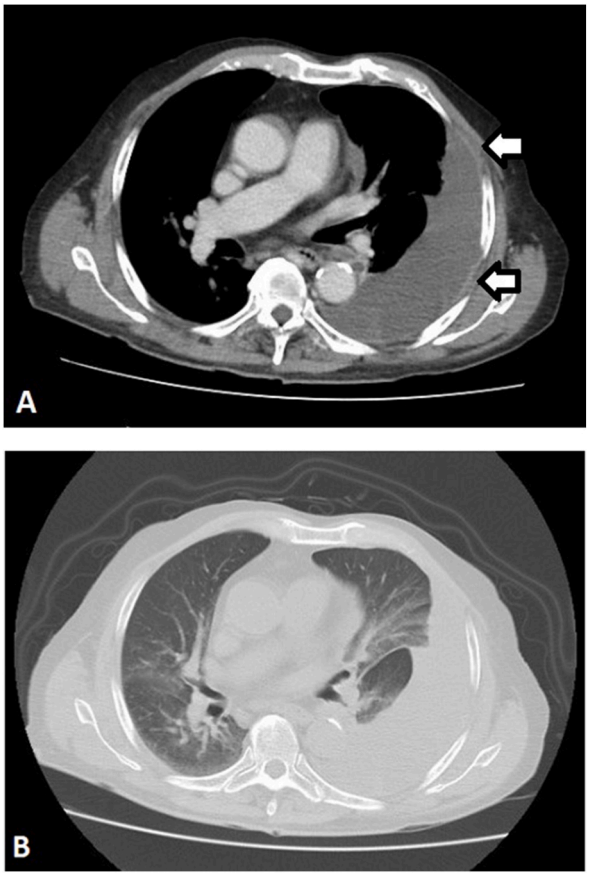

Fig. 1. Chest computed tomography scan. (A) Left pleural effusion with mild pleural thickening (arrow). (B) No significant parenchymal abnormalities are present.

nervous system, resulting in multiple episodes of seizures, hydrocephalus, and increased intracranial pressure resulting in headache, visual impairment, and vomiting.

Cysticerci may also invade the lung parenchyma and/or pleura, leading to pulmonary nodules, pneumonitis, lung cavities, or pleural effusion. Pleural cysticercosis is diagnosed by the demonstration of cysticercus within pleural tissue. Serum immunoglobulin G antibodies for cysticercosis can be used as a serological diagnostic test. Blood eosinophilia (absolute eosinophil count of >500/μL) is also useful evidence of disseminated disease. Wang et al. [7] reported that peripheral blood eosinophilia was found in 45.5 % of patients with pleural parasitic infestation. Eosinophilic pleural effusion (≥10 % eosinophils within pleural fluid) can also serve as evidence of parasitic infection. Previous case reports have demonstrated eosinophilic pleural effusion in most patients with pleural cysticercosis [1,2,8]. However, because this sign is nonspecific, other conditions characterized by eosinophilic pleural effusion must also be considered (e.g., hemothorax, malignancy, and tuberculosis).

Our patient presented with subacute symptomatic left pleural effusion causing shortness of breath, dry cough, pleuritic chest pain, and loss of appetite. Malignant pleural effusion was a differential diagnosis; however, the patient's pleuritic chest pain was not associated with the typical clinical manifestation of malignant pleural effusion. In the series of Marel et al., patients with malignant pleural effusions are more likely to have dull chest pain (34 % vs. 11 %), whereas patients with benign disease are more likely to have pleuritic chest pain (51 % vs. 24 %) [9]. Our patient had no peripheral blood eosinophilia. Pleural fluid analysis revealed a grossly bloody appearance and eosinophilic pleural effusion, but a definitive diagnosis was not obtained. Thus, the patient was advised to undergo pleuroscopy for a definitive diagnosis. This examination (Fig. 2) revealed diffuse pleural thickening with areas of inflammation. Interestingly, several small to large pleural nodules (up to 2 cm in diameter) were found in the posterior part of the parietal pleura with caudal predominance. Unlike a malignancy, the nodules were discrete, smooth-surfaced, round, and purplish, and no plaque-like lesions were found. Pathological examination of the biopsied nodules demonstrated a fibrotic cyst wall with a degenerated scolex, consistent with cysticercosis.

Few case reports on pleural cysticercosis have been published; however, other forms of pulmonary involvement have been described, including nodules, lung cavities, and patchy infiltrates on CT [1,2]. A typical chest CT finding is the presence of multiple variably sized pulmonary nodules [10]. Our patient initially underwent chest CT to examine his lung parenchyma after thoracentesis. The results revealed moderate left pleural effusion without other abnormalities (Fig. 1). Prior reports have indicated that chest pain or tightness is the most common respiratory symptom of pulmonary cysticercosis manifesting as pleural effusion [1,2], consistent with

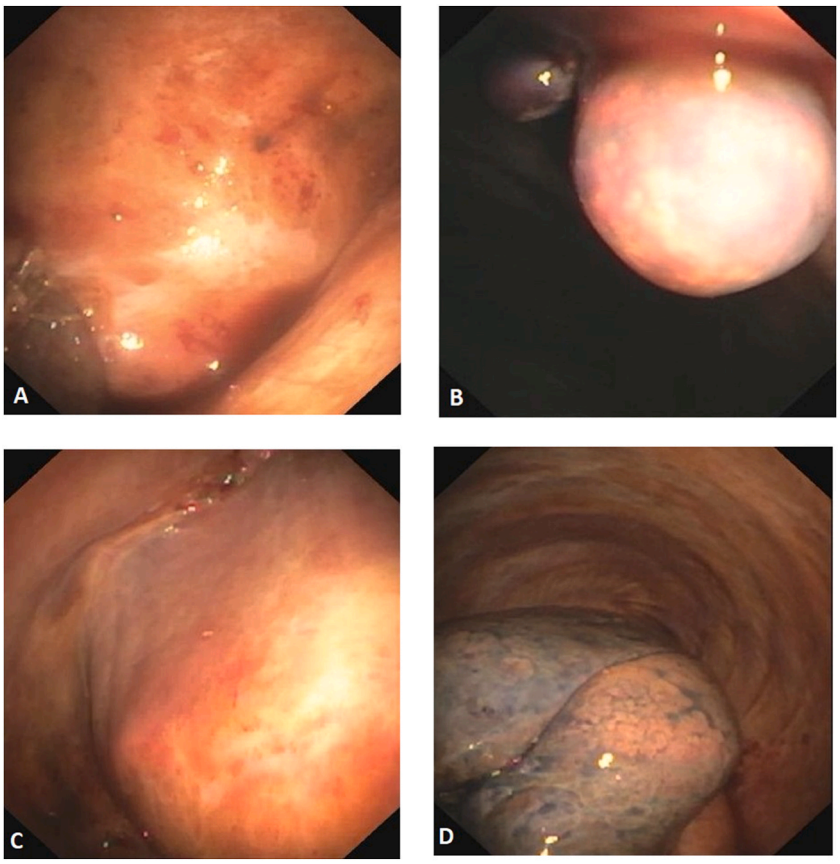

Fig. 2. Pleuroscopic findings. (A) Thickened and inflamed parietal pleura. (B) Cysticercal pleural nodules at posterior part of parietal pleura. (C) Diaphragm. (D) Apex.

Table 1
Pleural fluid analysis & closed pleural biopsy results.

Appearance	Bloody
pH	7.439
Glucose (mg/dL)	104
Total protein (g/dL)	4.4
Lactate dehydrogenase (U/L)	1606
RBC (/μL)	930,000
WBC (/μL)	2730
- Neutrophils (%)	24
- Lymphocytes (%)	51
- Eosinophils (%)	25
Adenosine deaminase (U/L)	24.7
Pleural fluid cytology	Negative for malignancy
Closed pleural biopsy	- Chronic pleuritis - No evidence of granuloma or malignancy

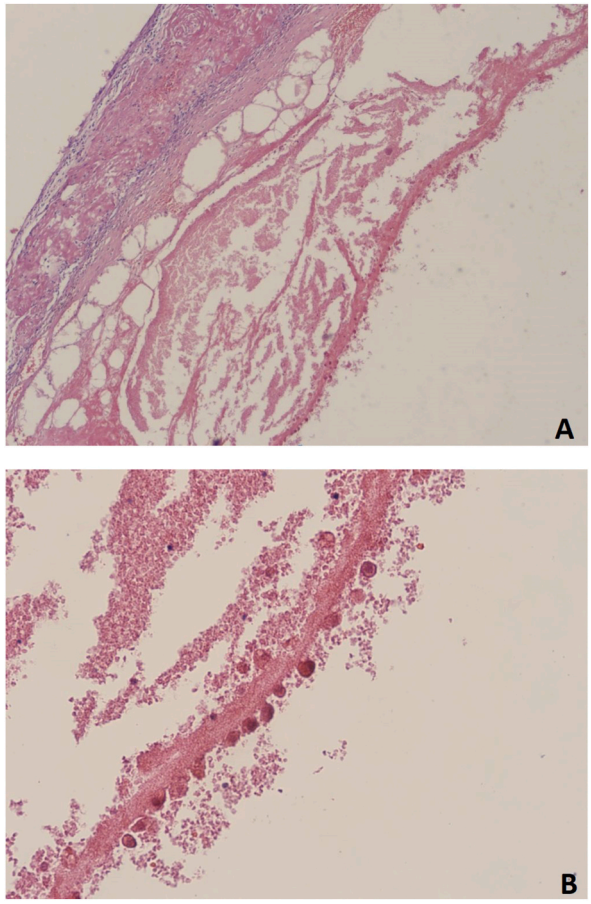

Fig. 3. Photomicrographs. (A) Degenerated cyst wall of cysticercus with eosinophilic outer cuticles. No protoscolex can be discerned (hematoxylin and eosin stain, original magnification × 40). (B) Acellular homogeneous eosinophilic membrane (hematoxylin and eosin stain, original magnification × 100).

our case. After being diagnosed with cysticercosis, our patient underwent further examination of his vital organs, including the brain and eyes, and no abnormalities were found. Albendazole (15 mg/kg/day) or praziquantel (50–60 mg/kg/day) have been prescribed for pulmonary cysticercosis in previous studies [1–4]. Thus, our patient received both albendazole and praziquantel for 2 weeks and showed a good response to treatment. Oral prednisolone was also prescribed to reduce the inflammatory response to the dead parasites.

To the best of our knowledge, this is the first report of a patient with isolated pleural cysticercosis without involvement of other sites. The appearance of cysticercus-induced pleural nodules was a novel pleuroscopic finding in this case. Although rare, pleural

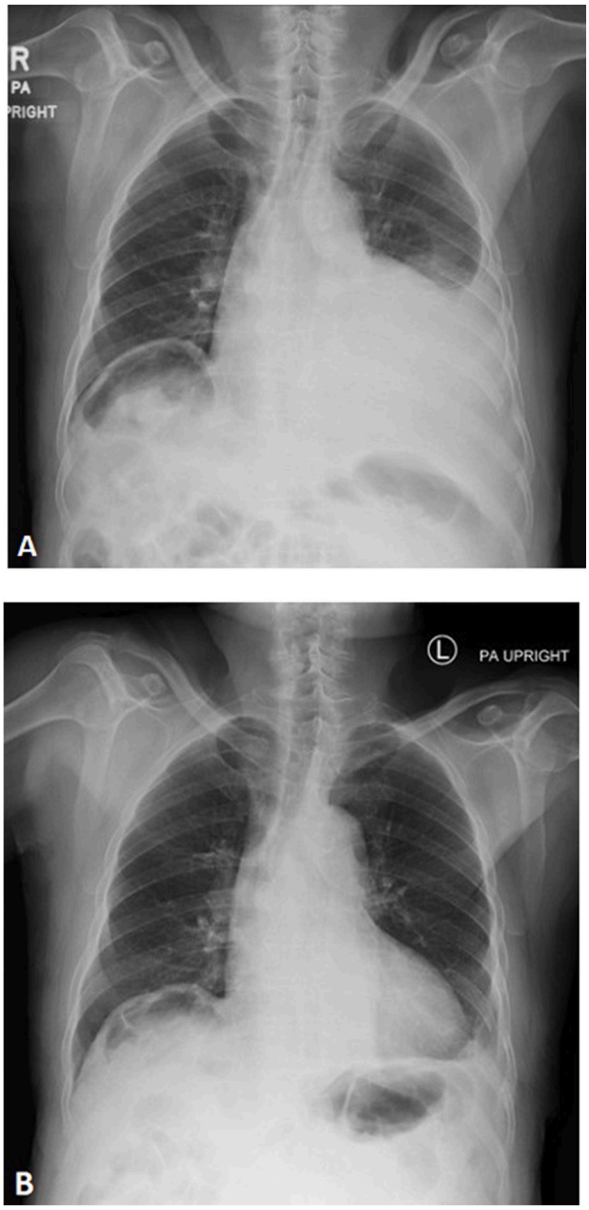

Fig. 4. Chest radiographs. (A) Before treatment. (B) After 4 weeks of treatment.

cysticercosis should be considered in patients with subacute symptomatic eosinophilic pleural effusion who have behavioral risk factors for cysticercosis, especially those living in endemic areas.

CRediT authorship contribution statement

Pornchai Opartpunyasarn: Writing – original draft, Investigation, Data curation, Conceptualization. **Sumeth Termmathurapoj:** Writing – review & editing, Supervision, Investigation. **Anan Wattanathum:** Writing – review & editing, Supervision, Investigation.

Ethics statement

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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