Calcified Pleural Tumor as a Rare Presentation of Paragonimiasis: A Case Report

Chi-Hsien Chen, Jih-Shuin Jerng, Pei-Shin Huang*, Pan-Chyr Yang

Calcified pleural tumor is a rare presentation in paragonimiasis, a parasitic disease that is endemic in Taiwan. The lung and pleura are the most common destinations for the parasite. Pleuropulmonary manifestations have been widely reported, and may mimic several lung diseases, such as lung cancer and pulmonary tuberculosis. The diagnostic requirement is a search for ova in the pleural effusion or pleural tissue. We report a 62-year-old woman who had suffered from intermittent right upper quadrant pain for 7 years. Computed tomography of the chest showed an 8-cm centrally calcified mass lesion in the right costophrenic angle. Biopsy of the mass revealed *Paragonimus westermani* ova surrounded by dense hyalinized fibrotic tissue. This case demonstrates that paragonimiasis may present as a calcified pleural tumor. ***(Thorac Med 2008; 23: 25-30)***

Key words: paragonimiasis, *Paragonimus*, pleural, calcification, tumor, abdominal pain

Introduction

Paragonimiasis, a parasitic disease that often causes pleuropulmonary problems, is endemic in the Asia-Pacific area [1]. Although paragonimiasis can be found worldwide, special methods of food preparation in the Asia-Pacific region promote human transmission. In the life cycle of *Paragonimus*, the ingested metacercariae excyst in the gastrointestinal tract, developing into larvae that penetrate the gut, peritoneum, diaphragm, and pleura into the lung. A pleuropulmonary lesion is the most common manifestation of *Paragonimus* infection [1]; its presentation may mimic diseases such as tuberculosis, fungal infection, and malignancy [1-3]. Calcified pleural tumor, however, is a rare presentation of paragonimiasis. Herein, we describe a patient with pleural paragonimiasis presenting as a calcified pleural tumor initially diagnosed as a chronic post-traumatic intra-pleural hematoma.

Case Report

A 62-year-old woman visited the hospital because of an intermittent dull pain in the right upper costal region after a traffic accident 7 years earlier. The patient recalled a severe bump on her right chest cage shortly after the accident. Chest radiography a month after the accident revealed an 8-cm mass-like lesion with extensive calcification in the right pleural cavity (Figure

Department of Internal Medicine, National Taiwan University Hospital, Taipei, Taiwan, *Department of Pathology, National Taiwan University Hospital, Taipei, Taiwan
Address reprint requests to: Dr. Jih-Shuin Jerng, Department of Internal Medicine, National Taiwan University Hospital, No. 7 Chung-Shan South Road, Taipei 100, Taiwan

1). Post-traumatic intrapleural hematoma was suspected. The pain could be relieved by analgesics, but would recur. Tc-99m DISIDA imaging showed dyskinesia of the gall bladder. Progressive enlargement of the lesion in the non-calcified part was noticed on the annual follow-up radiographs. There was no fever, chills, night sweats or weight loss.

On admission, her temperature was 36.5ºC, pulse rate was 72 beats per minute, and respiratory rate was 17 breaths per minute. The patient's blood pressure was 122/72 mmHg and the oxygen saturation was 98% while breathing ambient air. On physical examination, scoliosis was noted, the breathing sound was clear, and there was no chest wall deformity, no lymphadenopathy, and no abdominal tenderness. The hemogram revealed a red blood cell count of 4.41 M/μL, hemoglobin of 13.6 g/dl, platelet count of 193 K/μL and white blood cell count of 5.49 K/μL, with 57.3% polymorphonuclear leukocytes, 2.6% eosinophils, and 33.4% lymphocytes. The results of her blood biochemistry were as follows: albumin, 4.4 g/dl; total protein, 7.1 g/dl; blood urea nitrogen, 12.6 mg/dl; serum creatinine, 0.8 mg/dl; sodium, 141 mmol/L; potassium, 3.6 mmol/L; calcium, 2.2 mmol/L; asparate aminotransferase (AST), 20 U/L; alanine aminotransferase (ALT), 13 U/L; total bilirubin, 0.88 mg/dl; alkaline phosphate (ALP), 112 U/L; gama glutamyl transpeptidase (GGT), 22 U/L; lactate dehydrogenase, 502 U/L, and C-reactive protein, 0.09 mg/dl. Computed tomography showed an 8-cm, contrast-enhanced pleural mass located above the right costophrenic angle, with central calcification and thickened adjacent pleura (Figure 2A, 2B), and an equivocal connection with the twelfth rib (Figure 2C); there was a tiny nodule in the right middle lung (Figure 2D). CT guided biopsy of the mass revealed dense hyaline fibrotic tissues with opercular *P. westermani* ova within the necrotic debris (Figure 3), compatible with paragonimiasis. Surgical intervention was not recommended, and the patient was treated with praziquantel 600 mg daily for 3 days. In the 5-month follow-up, the costal pain had subsided completely, despite the mass size remaining unchanged. The patient was then followed up regularly.

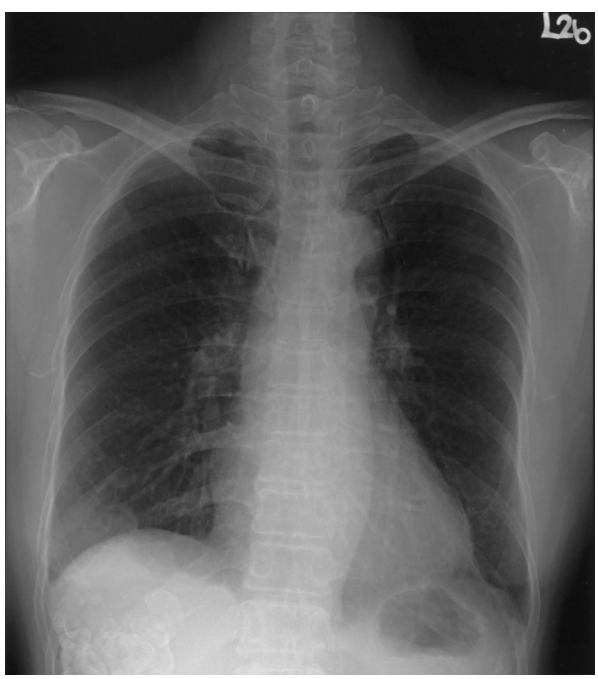

Fig. 1. Chest radiography showing an 8-cm mass-like lesion with extensive calcification in the right basal pleural cavity

Tracing back her history, she was brought up in Hsinchu County. Freshwater mitten crab has been a popular dish in her family and in that area. She denied having eaten raw crabs in her childhood, but incompletely-cooked crabs or contamination during the cooking process should be considered as possible causes of her parasitic lung disease. She also recalled that her cousin, who lived with her family at that time, had been found to have lung paragonimiasis when she was 7 years old. Her family has not cooked freshwater mitten crabs since then.

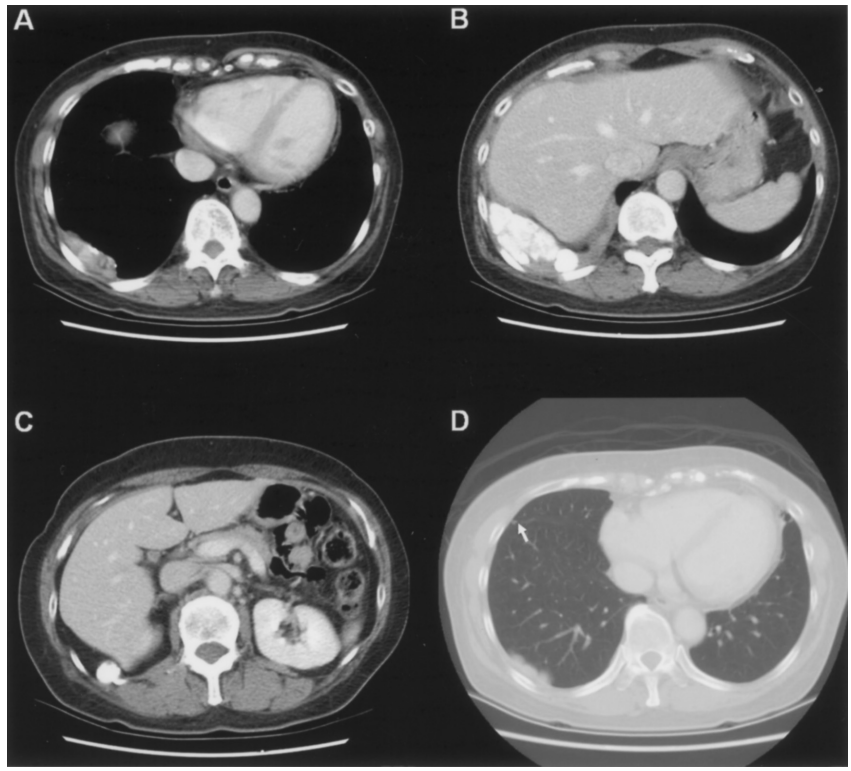

Fig. 2. Chest computed tomography in the mediastinal window. (A) An 8-cm, contrast-enhanced pleural mass located above the right costophrenic angle, with (B) central calcification and thickened adjacent pleura, and (C) an equivocal connection to the 12th rib. (D) In the lung window, a tiny nodule in the right middle lung can be seen (arrow).

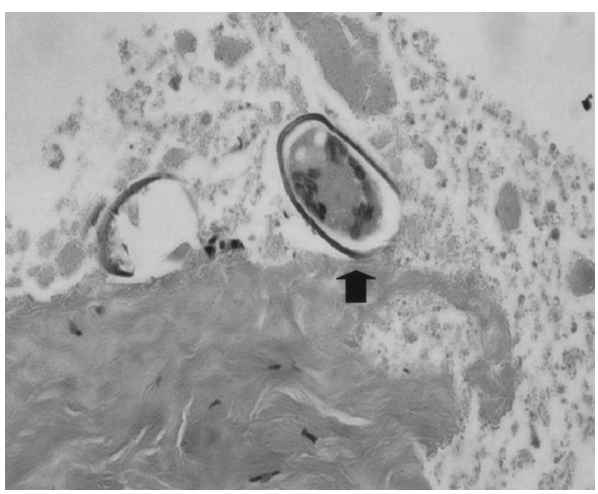

Fig. 3. CT guided biopsy specimen showing dense hyalinized fibrotic tissue admixed with *P. westermani* ova (arrow). Hematoxylin and eosin stain, original magnification, 200×.

Discussion

Humans are one of the definitive hosts of *P. westermani*. Ingestion of metacercaria, the infectious form of *Paragonimus* fostered in freshwater crab or crayfish, may lead to permanent mammalian infection. Yokogawa *et al.* has reported that raw crab juice left on the knife or chopping block might contaminate other foods, which then become another infectious route for lung fluke [1]. The larvae might migrate across the diaphragm into the pleural space and enter the lung, where they develop into adult flukes. [1, 4] The mass lesion found at the costophrenic angle in this reported case is explained by their usual migrating route.

The exact life span of *P. westermani* in human beings is not completely understood, but the average life span is usually less than 10 years. There have been reports showing that eggs of *Paragonimus westermani* could be found in sputum 30 years after leaving the endemic area [1]. To our knowledge, this case had the longest latent period (probably more than 50 years) that has been reported in the literature. The calcification may be evidence of a long-term process. However, reinfection cannot be completely ruled out in an endemic area.

The clinical presentation of paragonimiasis depends on the load of larvae and the stage of their migration. Most patients at the early stage experience pleuritic chest pain due to pleurisy. Pleural effusion, hydropneumothorax, or pleural thickening may be found in pleural paragonimiasis [1, 3-7]. After the flukes enter the lung, cough and hemoptysis become the major symptoms [1, 7]. Pulmonary paragonimiasis usually present with a nodule, cyst or mass-like lesion located in the peripheral lung in radiographic studies. In this patient, the nodule in the right middle lung might be considered a pulmonary involvement of paragonimiasis. Although the above presentations may be nonspecific [2, 6], some signs on images may point to the diagnosis of paragonimiasis. A linear opacity in the lung, which suggests a worm migration tract, or a combination of localized pleural thickening and subpleural linear opacities connecting pleura and a necrotic lung nodule were all typical findings of paragonimiasis in chest computed tomography [3, 8].

The differential diagnoses for a calcified pleural tumor might include mesothelioma [9-11], chondrosarcoma, osterosarcoma, ganglioneuroblastoma [12-13], chronic tuberculous or bacterial empyema with fibrothorax [14-15], chronic hematoma [16], and fibrous pseudotumor of the pleura [17]. This patient had a history of traumatic injury, and recent hematoma, which could also mimic a centrally calcified pleural tumor with diffuse hyperdensity, had once been considered [16]. The attenuation of hematoma on CT scans is mainly contributed to by hemoglobin, and a linear relationship between attenuation and hematocrit has been established [16, 18]. Usually, a search for ova from pleural effusion or biopsy tissue is needed to confirm the diagnosis of paragonimiasis.

Paragonimus may also invade other organs such as the brain, liver, kidney, peritoneum and spinal cord [1], and calcification in the lung, brain, skeletal muscle, and liver has been reported [7, 15]. To our knowledge, paragonimiasis presenting with a calcified pleural mass has never been reported.

Treatment for paragonimiasis with praziquantel has been effective [19-20], but in some patients, bronchiectasis or atelectasis do not recover [7]. The only indication for surgical intervention in this disease is decortication for chronic empyema [7, 21], which was not present in this reported case.

In summary, this case of paragonimiasis with a rare presentation of an extensively calcified pleural mass may alert clinicians to the importance of taking *P. westermani* infection into consideration in patients with chronic calcified pleural lesions.

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以鈣化性肋膜腔腫瘤爲表現的肺吸蟲症：一病例報告

陳啟信　鄭之勛　黃佩欣 *　楊泮池

臺灣地處於肺吸蟲症流行病區，和食用未經熟煮的淡水蝦蟹有關。人類是肺吸蟲的宿主之一，食入的幼蟲會穿過腸道，腹膜，橫隔膜，和肋膜而進入肺臟。因此肺臟和肋膜腔內的變化是肺吸蟲最常見的臨床表現。過去的研究報告顯示，肺吸蟲在肺臟和肋膜腔的表現可以和肺結核與惡性腫瘤相似。而診斷的確立須要經由蟲卵的發現。然而就我們所知，以鈣化的肋膜腔內腫瘤來表現的肺吸蟲症並未被報導過。本個案指出，肺吸蟲症有可以靠近橫隔膜的鈣化腫瘤來表現，且合併長期的右上腹疼痛。*(胸腔醫學* **2008; 23: 25-30)**

關鍵詞：肺吸蟲病，肺吸蟲，胸膜的，鈣化，腫瘤，腹痛

台大醫院　內科部，* 台大醫院　病理部
索取抽印本請聯絡：鄭之勛醫師，台大醫院內科部，台北市中山南路 7 號