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Chest

Castleman's disease presenting as a pleural tumor: a case report with CT findings

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ABSTRACT

Castleman's disease (CD) is an uncommon benign lymphoproliferative disorder which most commonly involves the mediastinum but rarely affects the pleura. We report a case of unicentric CD that presents as a pleural mass in a 45-year-old man, which was subsequently resected followed by an unexpected diagnosis on histologic examination. Although rare, CD should be included in the differential diagnosis of well-enhancing pleural mass.

Introduction

Castleman's disease (CD) is a rare, benign lymphoproliferative disorder. It can occur most commonly in the mediastinum, but other areas including neck, abdomen, and axilla are common sites as well. CD arising from the pleura is rare [1].

In this case, we describe the rare presentation of unicentric CD, which presented as a pleural mass mimicking other tumors in the pleura or chest wall.

Case report

A 45-year-old man presented with dyspnea for 5 weeks and 10 kg of weight loss during 1 month. He had no significant medical history and initial laboratory workup revealed HIV negative.

Chest computed tomography (CT) demonstrated a well-defined, ovoid soft tissue mass in the left lower hemithorax, about 4.5 × 3.5 cm in size. The lesion had a broad base toward

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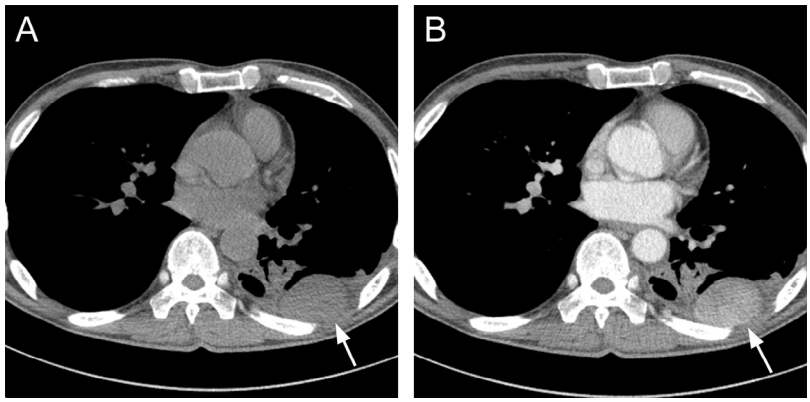

Fig. 1 – Contrast-enhanced CT of pleural Castleman's disease. (A) Precontrast CT scan shows a pleural-based mass lesion (arrow) in the posterior left hemithorax at the level of the 8th rib measuring approximately 4.5 × 3.5 cm in axial dimensions. (B) Contrast-enhanced image shows homogenous avid enhancement of the mass. CT, computed tomography.

the pleura. The mass was homogenously well-enhancing, which reflected its hypervascular nature (Fig. 1). The radiological differential diagnoses of the mass were neurogenic tumor or solitary fibrous tumor of the pleura. Additionally, CT showed moderate amount of loculated fluid collection with internal air foci and pleural thickening with enhancement in the left lower hemithorax. Pleural fluid examination showed pH 7.5, glucose 7 mg/dL, protein 4.3 g/dL, white blood cell 15,750/mm^3 (polymorphonuclear leukocyte 70%, lymphocyte 30%). The patient underwent percutaneous catheter drainage for empyema and administrated intravenous antibiotics. His clinical symptoms and chest radiograph findings improved.

For the diagnosis of the mass in the left hemithorax, CT-guided core needle biopsy of the lesion was performed. Microscopic findings revealed dense lymphoplasmacytic proliferation, suggesting chronic inflammation or hematolymphoid disease. In particular, 1 lymphoid follicle with a penetrating capillary was noted, which is one of the findings of CD.

The patient was scheduled for thoracoscopy for further evaluation. On thoracoscopy, the lesion was found to be a well-

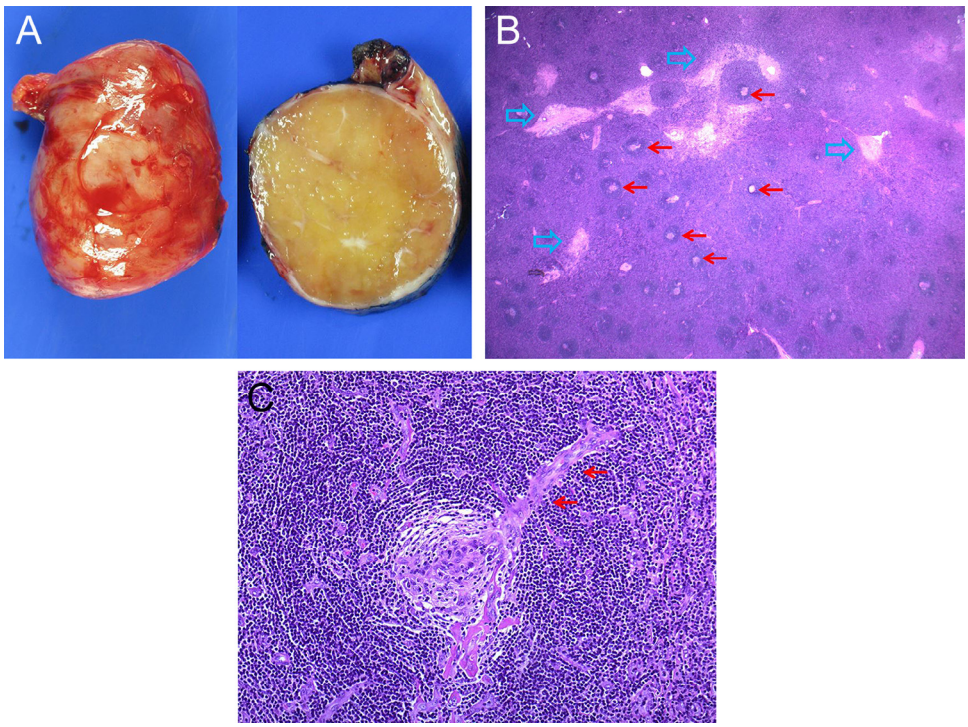

Fig. 2 – Histopathologic findings of pleural Castleman's disease. (A) Macroscopy of the excised tumor demonstrates well-encapsulated mass with yellow-tan color without hemorrhage or necrosis. (B) Numerous lymphoid follicles (red arrows) were noted throughout the cortex and medulla. Sclerotic stroma (blue open arrows) was also found (hematoxylin and eosin ×12.5). (C) The lymphoid follicle was partially lymphocyte-depleted and penetrated by a blood vessel (red arrows) (hematoxylin and eosin ×200).

defined mass, arising from the posterior parietal pleura. As severe adhesion was noted due to empyema; thoracotomy was performed, and the lesion was resected completely. The mass lesion was well-encapsulated. The cut surface showed a homogenous tan-yellow color with white fibrotic areas. No hemorrhage or necrosis was detected. On histologic examination, low magnification power showed dense lymphoid proliferation with prominent follicles. Some of them were involute with hyalinization and some had penetrating capillaries. Subcapsular sinus was absent. Widened mantle zone showed concentric layering of lymphocytes. Vascular proliferation was noted in the interfollicular area. The vessels had hyalinized walls. The pathologic findings were consistent with a hyaline vascular variant of CD (Fig. 2).

Discussion

CD is an uncommon lymphoproliferative disorder, which was first described in 1956 by Benjamin Castleman [2]. It can involve any site with lymphatic tissue throughout the body, and most commonly presents in the mediastinum, neck, retroperitoneum, and axilla [3]. Uncommonly, it may arise at other locations, including the lung, trachea, pleura, and pericardium.

Clinically, it may manifest as 2 major subgroups: unicentric localized disease and multicentric systemic disease, and histologically it is classified as 2 major subtypes: hyaline-vascular and plasma-cell [4,5]. The hyaline-vascular variant accounts for the majority of unicentric disease and is characterized by lymphoid follicular hyperplasia and vascular proliferation into the interfollicular region [5]. Enlarged lymph nodes that demonstrate intense homogeneous contrast enhancement reflecting the hypervascularity of the lesion are the most common CT finding of a hyaline vascular variant of CD [6,7]. This intense enhancement, which helps narrow the differential diagnosis, is secondary to the presence of an extensive network of small blood vessels in the interfollicular zone of the hyaline vascular subtype, which represents the vast majority of unicentric CD (UCD) cases [4].

A few cases of CD of the pleura have been reported with imaging, and the largest series consists of 8 patients reported by Ko et al. [1]. Most of the patients were asymptomatic and the disease was incidentally detected. In our case, the patient presented with empyema and the pleural mass was detected incidentally. It could mimic both benign and malignant conditions in the chest wall or pleura. The main differential diagnosis of tumors in the posterior mediastinum or pleural space includes neurogenic tumors and solitary fibrous tumor. Differentiation between these lesions and UCD at CT is difficult, but UCD tends to demonstrate a more pronounced enhancement than these other lesions [8]. Although intense contrast enhancement of the lesion was shown in our case, it was difficult to diagnose CD without any histologic findings due to its unusual location.

CD can occur at any site with a lymphatic tissue. Mass or enlarged lymph nodes that demonstrate intense homogeneous contrast enhancement on CT might be an implication of CD. Although establishing an accurate preoperative diagnosis is difficult, our case suggests that CD can be included in the differential diagnosis of thoracic lesion with distinct margin and obvious enhancement on CT even when located in the chest wall or pleura.

References

[1] Ko SF, Ng SH, Hsieh MJ, Lin JW, Huang CC, Lee TY, et al. Castleman's disease of the pleura: experience with eight surgically proven cases. Ann Thorac Surg 2003;76:219–24.
[2] Castleman BTV. Case records of the Massachusetts General Hospital weekly clinicopathological exercises: case 40011. N Engl J Med 1954;250:26–30.
[3] Soumerai JD, Sohani AR, Abramson JS. Diagnosis and management of Castleman disease. Cancer Control 2014;21(4):266–78.
[4] McAdams HP, Rosado-de-Christenson M, Fishback NF, Templeton PA. Castleman disease of the thorax: radiologic features with clinical and histopathologic correlation. Radiology 1998;209(1):221–8.
[5] Cronin DM, Warnke RA. Castleman disease: an update on classification and the spectrum of associated lesions. Adv Anat Pathol 2009;16(4):236–46.
[6] Ko SF, Hsieh MJ, Ng SH, Lin JW, Wan YL, Lee TY, et al. Imaging spectrum of Castleman's disease. AJR Am J Roentgenol 2004;182(3):769–75.
[7] Bonekamp D, Horton KM, Hruban RH, Fishman EK. Castleman disease: the great mimic. Radiographics 2011;31(6):1793–807.
[8] Kligerman SJ, Auerbach A, Franks TJ, Galvin JR. Castleman disease of the thorax: clinical, radiologic, and pathologic correlation: from the radiologic pathology archives. Radiographics 2016;36(5):1309–32.