—Case Reports—

Mediastinal Cystic Hemangioma Presenting as Bilateral Bloody Pleural Effusion: A Case Report

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Abstract

A 58-year-old man was admitted to our hospital because of dyspnea, dysphagia, and back pain. A chest roentgenogram showed bilateral pleural effusion. Bloody fluid was aspirated via thoracocentesis, but no malignancy was detected on cytological examination. Computed tomography revealed a hypodense circular mass approximately 8 cm in diameter with well-defined margins in the posterior mediastinum. Furthermore, T2-weighted magnetic resonance imaging showed the lesion to be a hyperintense mass. Video-assisted thoracoscopic surgery was performed, and the tumor was diagnosed as a mediastinal cystic hemangioma. The bilateral bloody pleural effusion was attributed to the cystic tumor.
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Key words: mediastinal tumor, hemangioma, cystic hemangioma, bloody pleural effusion

Introduction

Mediastinal hemangiomas are rare neoplasms, and cystic lesions are extremely rare. This report describes a patient presenting with bilateral bloody pleural effusion. A cystic hemangioma of the posterior mediastinum was diagnosed after it was surgically resected via video-assisted thoracic surgery.

Case

A 58-year-old man was admitted to our hospital because of dyspnea, dysphagia, and back pain. A chest roentgenogram showed bilateral pleural effusion (**Fig. 1**). Approximately 1,600 mL of bloody fluid was aspirated from the left hemithorax via thoracocentesis. Cytological examination showed no malignancy, and no microorganisms (including those causing tuberculosis) were detected in the aspirated pleural effusion. Laboratory data, including levels of tumor markers (carcinoembryonic antigen, cytokeratin fragment, and neuron-specific enolase), were all normal except the hemoglobin concentration (8.5 g/dL), which indicated anemia.

Computed tomography (CT) revealed a hypodense, markedly expanded mass approximately 8 cm in diameter in the posterior mediastinum (**Fig. 2A**). No abnormality was found in the lung fields or

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the parietal pleura. T2-weighted magnetic resonance imaging (MRI) showed the lesion to be a 10- × 8-cm hyperintense mass **(Fig. 2B)**. ^{18}F-fluorodeoxy glucose positron-emission tomography (FDG-PET) was performed to determine if the mass was malignant; however, the lesion did not show any uptake (data not shown). In the 2 months after admission, 4,000 mL of pleural effusion was aspirated bilaterally via repeated thoracocenteses.

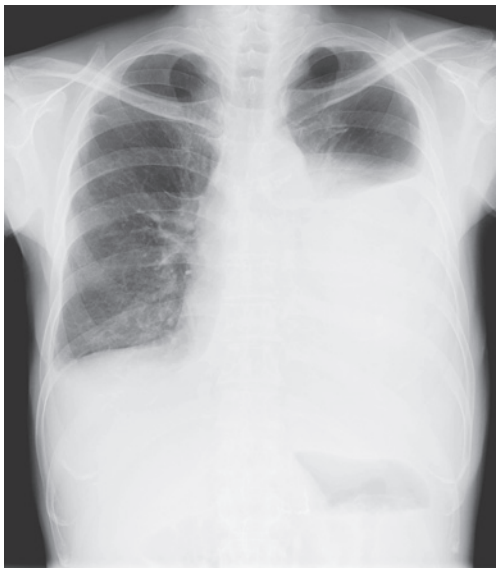

Fig. 1 Chest roentgenogram on admission showed bilateral pleural effusion, particularly in the left thorax.

Video-assisted thoracic surgery was performed to resect the cystic tumor. Under combined general and epidural anesthesia with a double-lumen endotracheal tube, the patient was placed in the right lateral decubitus position. First, a 5-mm port was inserted in the left seventh intercostal space at the midaxillary line, and the intrathoracic condition was observed through a thoracoscope. Little pleural effusion was observed in the left thoracic space because thoracocentesis had been performed 2 days earlier. Next, a minithoracotomy (8 cm in length) was made in the left ninth intercostal space starting at the posterior axillary line. The cyst surrounded the pleural ligament, as had been observed on CT and MRI **(Fig. 3A)**. The cyst wall was resected from the bottom of the cyst (just above the diaphragm) with an ultrasonic cutting and coagulation device **(Fig. 3 B)**. The cyst had a thick wall and was filled with fluid containing blood. After the cyst wall had been resected as completely as possible, the residual cyst was cauterized from the inside with a low-output electrocautery device. The patient's postoperative recovery was excellent, and the thoracic drain was removed 2 days after surgery.

Histological examination revealed that the cyst wall consisted of elastic fibers, and some layers of the endothelium of the hemangioma wall were stained for CD34 and showed inflammatory cell

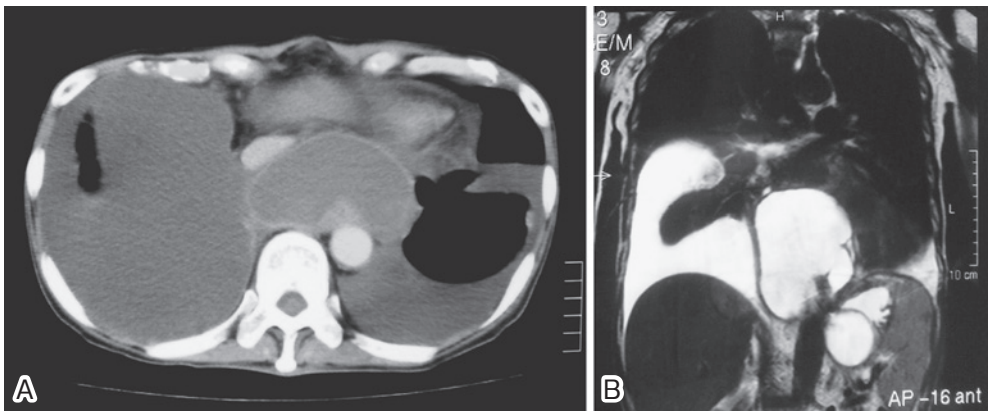

Fig. 2 **A**: Chest computed tomography revealed a markedly expanded hypodense mass approximately 8 cm in diameter with bilateral pleural effusion in the posterior mediastinum. The cyst was observed to compress the lower part of the heart towards the anterior chest wall.
B: T2-weighted magnetic resonance imaging of the chest showed a hyperintense lesion, similar in intensity to pleural effusion. The 8- × 10-cm mass was in the posterior mediastinum.

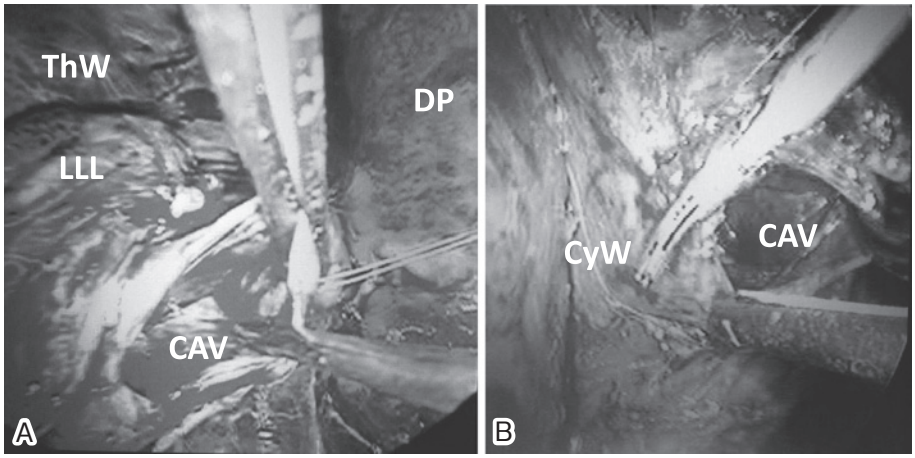

Fig. 3 **A, B**: The cyst, located between the left lower lobe and diaphragm, had a thick wall and was filled with bloody fluid. The cyst wall was resected from its bottom by means of an ultrasonic cutting and coagulation device.
ThW, thoracic wall ; LLL, left lower lobe ; DP, diaphragm ; CAV, cavity ; CyW, cyst wall

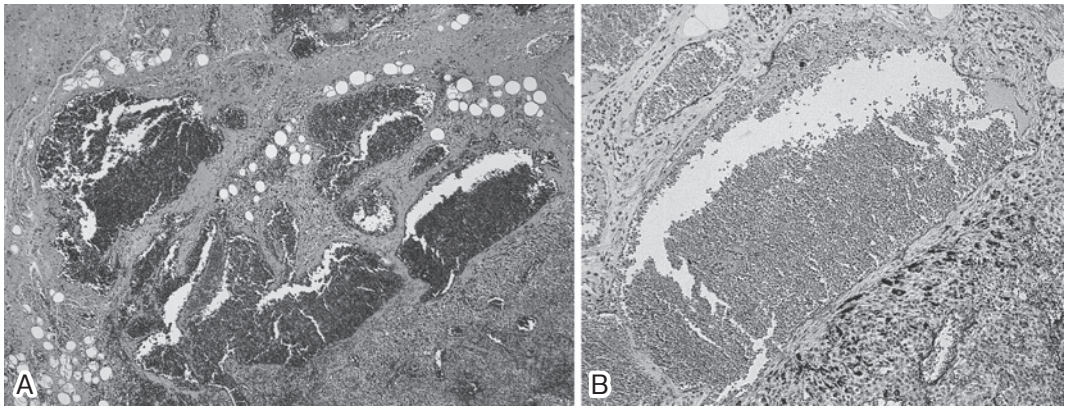

Fig. 4 **A, B**: Hematoxylin and eosin and anti-CD34 staining of the cyst wall revealed the presence of elastic fibers. Some layers of the endothelium of the hemangioma wall were stained for CD34.

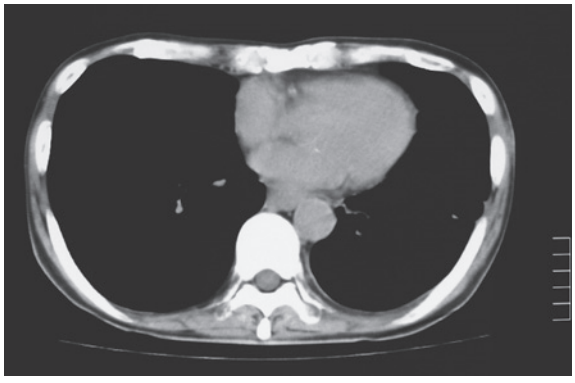

Fig. 5 Chest computed tomography (1 year after surgery) showed no pleural effusion and no cystic lesion in the thorax.

infiltration **(Fig. 4A, B)**. Staining for D2-40 to detect lymphoid tissues was negative (data not shown). Finally, the cystic tumor was diagnosed as a benign cystic hemangioma of the mediastinum. One year after surgery, chest CT showed no pleural effusion and no cystic lesion in the thorax **(Fig. 5)**.

Discussion

A mediastinal hemangioma is a rare neoplasm. In 1982, Wada and Teramatsu reported that hemangiomas accounted for 0.3% (5 of 1,546) of mediastinal tumors in Japan[1]. Furthermore, Gindhart et al have reported that hemangiomas comprise less than 0.5% of all mediastinal tumors[2]. Most

hemangiomas are located in the anterior mediastinum, and few have been reported in the posterior mediastinum[3]. Moreover, most of these hemangiomas are solid tumors, such as cavernous and capillary hemangiomas. Only 2 cases of cystic hemangiomas of the mediastinum have been reported in Japan[4,5], but these hemangiomas were in the anterior mediastinum. Hence, our case is an extremely rare case of a cystic hemangioma in the posterior mediastinum.

Preoperative CT and MRI suggested that this mediastinal tumor was a cystic lesion containing low-viscosity liquid, and cytological examination and FDG-PET suggested that the likelihood of malignancy was low.

The first purpose of surgery was to diagnose the tumor, while the second was to create a conduit to contain the liquid from the cyst to the thoracic cavity by resecting the cystic wall to reduce symptoms, such as dysphagia and back pain.

The cyst could not be completely resected, but the discharge causing pleural effusion was markedly reduced. The chest tube was removed 2 days after surgery, and no increase in pleural effusion was observed through successive chest roentgenograms.

According to Cohen et al.[6], although 6 of 15 cases of benign cystic tumors were incompletely resected, no progression of residual tumors was observed during the postoperative follow-up period. They suggested that surgical resection, even when incomplete, would reduce tumor volume and decompress surrounding organs. Our patient also had no recurrence of pleural effusion or regrowth of the cyst 1 year after surgery.

The pathological diagnosis was benign cystic hemangioma and was not expected before surgery. Although oozing of effusion from the cyst could not be confirmed through intraoperative or pathological findings, the oozing was assumed to be due to the effusion from inside the cyst wall.

Conclusion

We have documented a case of a posterior mediastinal cystic hemangioma in a 58-year-old man which presented as bilateral bloody pleural effusion. Although the cyst was incompletely resected, no recurrence has been observed for at least 1 year since surgery. Nevertheless, continuous observation for recurrence is required.

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