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**Case
Report****Venous Hemangioma of the Posterior Mediastinum**

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Here, we describe our experience in treating a patient with mediastinal hemangioma, a rare neoplasm. An abnormal shadow was noted in the thoracic region of a 54-year-old woman at a health checkup, and she was referred to our hospital. A neurogenic tumor was suspected based on the findings of the chest X-ray and computed tomography scan. Thoracoscopic tumorectomy was performed. The tumor surface was smooth with a reddish-dark reddish color, and capillary blood vessels showed marked growth around the tumor. The tumor was composed of medium or large blood vessels with a relatively thick vascular wall containing smooth muscle. On immunostaining, anti-CD34 antibody and Factor VIII were positive and D2-40 was negative. Based on these findings, the tumor was diagnosed as mediastinal venous hemangioma.

Keywords: hemangioma, posterior mediastinum

Introduction

Mediastinal hemangioma is a very rare neoplasm. In most studies of mediastinal masses, hemangiomas have an incidence of 0.5% or less.^{1,2)} Here, we describe our experience in treating a patient with such a tumor.

Case Report

An abnormal shadow was noted in the thoracic region of a 54-year-old woman at a health checkup, and she was referred to our hospital. On chest X-ray radiography, a tumor with a regular margin measuring 3 cm in diameter and protruding rightward was present in the apex of the

right lung near the mediastinum. On chest computed tomography (CT), a tumor with a regular margin and homogeneous content measuring 27 mm in diameter was present in contact with the vertebral body at the Th2/3 level (**Fig. 1**). Contrast CT could not be performed because the patient had a past medical history of bronchial asthma.

A neurogenic tumor was suspected based on the above findings, and surgery was selected. Thoracoscopic tumorectomy was performed. The tumor surface was smooth with a reddish-dark reddish color, and capillary blood vessels showed marked growth around the tumor. The tumor was composed of medium or larger blood vessels with a relatively thick vascular wall containing smooth muscle (**Fig. 2A**). On immunostaining, anti-CD34 antibody and Factor VIII were positive (**Fig. 2B**), and D2-40 was negative (**Fig. 2C**). Based on these findings, the tumor was diagnosed as venous hemangioma.

Discussion

Benign hemangiomas of the mediastinum are rare tumors with an incidence of 0.5% of mediastinal masses or less.^{1,2)} The first case was reported by Shannon in 1914.¹⁻⁴⁾ According to Taori et al., only 125 well-documented cases of mediastinal hemangioma had been reported in

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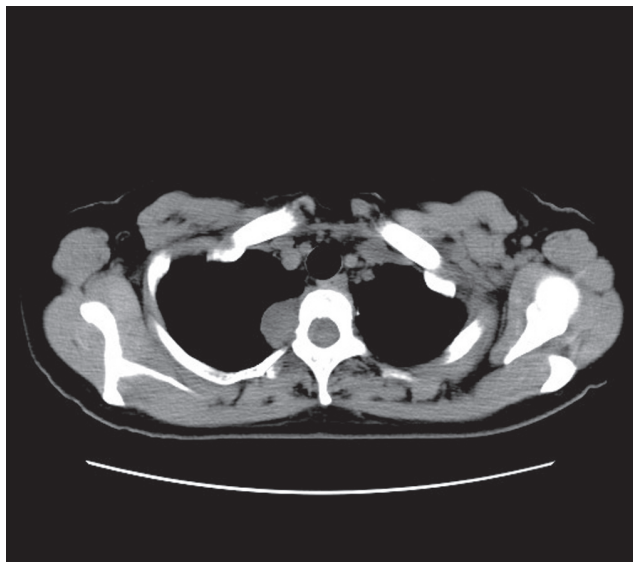
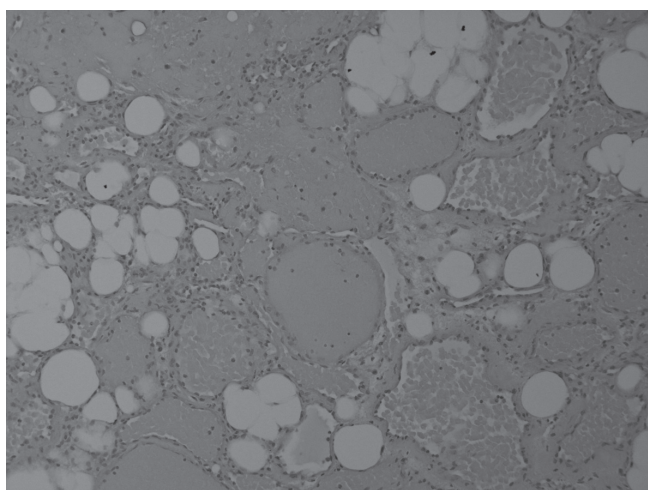
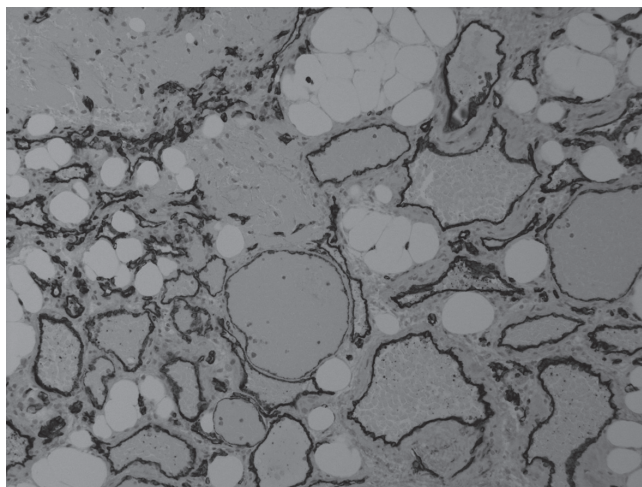
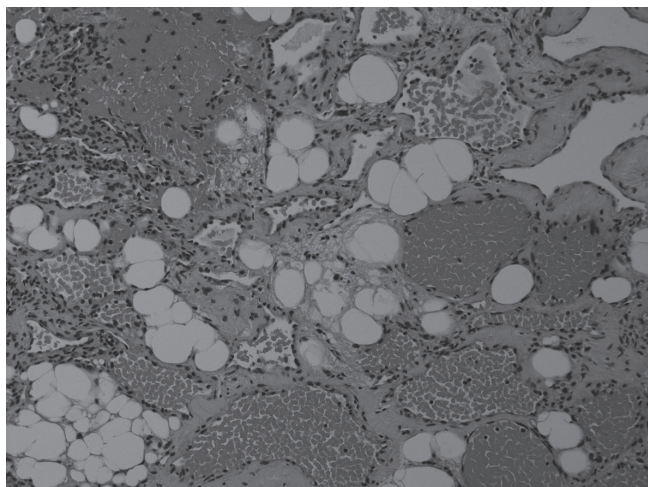


Fig. 1 On chest CT, a tumor with a regular margin and homogeneous content measuring 27 mm in diameter was present in contact with the vertebral body at the Th2/3 level.



A	B
C	

Fig. 2 Microscopic findings.

- A:** The tumor was composed of medium or larger blood vessels with a relatively thick vascular wall containing smooth muscle (HE $\times 20$).
- B:** One layer of the hemangio-endothelium was stained with anti-CD34 antibody.
- C:** Endothelial cells showed negative staining for D2-40, which specifically recognizes lymphatic endothelial cells.

the literature up until 2000.⁵⁾ There is a wide range of age at the time of diagnosis (26 days–76 years), but 75% of cases are first discovered at 35 years of age or younger, and the peak incidence is in the first decade of life.^{3, 6)} There is no sex predilection.^{3,7)} Hemangiomas are benign, richly vascular tumors that can involute with time. Histologically, hemangiomas appear as a proliferation of normal vascular elements and have various amounts of interposed stromal elements (eg, fat, myxoid, and fibrous tissue).⁸⁾ The tumors are categorized according to the size of their vascular spaces as capillary, cavernous, or venous hemangiomas.⁶⁾ The vast majority (90%) of cases were hemangiomas of either the cavernous or capillary type. The remainder were angiomas (2%), hemangiofibromas (2%), fibrogioma (1%), fibrolipohemangioma (1%), venous hemangioma (1%), and arteriovenous malformation (1%)³⁾; therefore, this case is relatively rare. In 68%, the anterior mediastinum was involved, and in 58%, it was the sole compartment affected. There were no instances of solitary involvement of the middle mediastinum. The posterior mediastinum was the primary site of involvement in 22% of cases.^{3,5)} This tumor arose in the posterior mediastinum, which is also rare. One-third to one-half of patients have no symptoms at presentation; the remainder present with non-specific symptoms, such as a cough, chest pain, and dyspnea due to compression or, less likely, invasion of adjacent structures.^{1,3,5,6)}

Mediastinal hemangiomas should be considered an important differential diagnosis of posterior mediastinal masses, even though these are rare.⁹⁾ The preoperative diagnosis may be difficult because the tumors usually manifest as nonspecific mediastinal masses on chest radiographs.^{2,6)} Hemangiomas appear as round or lobulated, well-defined masses. Calcified phleboliths are helpful in suggesting the vascular nature of the mass; however, phleboliths are seen in only 10% of cases.^{1,2,9,10)}

CT can demonstrate phleboliths more sensitively than conventional radiograph.⁸⁾ Most mediastinal hemangiomas manifested as well-margined masses at CT.⁶⁾ Cohen et al., in a retrospective surgical series, concluded that radiologic studies, including CT, had little to offer in the preoperative evaluation of these lesions.¹⁾ However, Seline et al. reported central “puddling” of contrast material within one mediastinal hemangioma on CT, a finding the authors believed might be diagnostic of mediastinal hemangioma.⁷⁾ The appearance of puddling of iodinated contrast material within the mass on a dynamic-enhanced CT using a bolus technique is probably diagnostic.⁷⁾ Many reports have stated that contrast CT reveals a slightly en-

hanced heterogeneous mass. According to McAdams et al. it is a characteristic of mediastinal hemangioma that the center of the tumor shows stronger enhancement than the margin on CT.²⁾

Magnetic resonance imaging (MRI) of the chest was approximately equal to that of the muscle on T1-weighted images, while it was distinctly higher than the muscle on T2-weighted images. Differences in signal intensity on T1- and T2-weighted images can be helpful for the diagnosis of mediastinal hemangioma. MRI with gadolinium-diethylenetriamine pentaacetic acid (Gd-DTPA) enhancement revealed a somewhat heterogeneous mass shadow of the tumor with marginal enhancement.²⁾ Sakurai et al. described that markedly high intensity on fat suppression T2-weighted image might be a characteristic finding.⁸⁾

Posterior mediastinal tumors are difficult to diagnose before surgery. Furthermore, the preoperative diagnosis of mediastinal hemangioma is also difficult. Currently, surgery is performed for the purpose of diagnosis and treatment. Even if the diagnosis of mediastinal hemangioma is made preoperatively, surgery will be selected to resolve tumor-related symptoms, but the procedure may be hazardous because of the potential complication of substantial blood loss.⁶⁾ In this case, thorascopic tumorectomy was performed, but blood flow was marked, unlike in general neurogenic tumors, and so blood loss was relatively high. Thorascopic excision of a hemangioma located in the anterior mediastinum has recently been reported.¹⁰⁾ Thorascopic tumorectomy of hemangioma may become increasingly selected in the future, and hemorrhage should be considered. When symptomatic massive hemangioma is present, surgery is considered. For surgery, the risk of hemorrhage must be considered. Even in patients with benign tumors, it is not necessary to adhere to thorascopic surgery, and surgery should be performed under thoracotomy in some patients. Although total resection is ideal, Cohen et al. reported that the results of subtotal excision were acceptable.¹⁾ When total resection is difficult, subtotal excision may be selected.

Anti-CD34 antibodies have been reported to stain blood vessels in tissue sections.⁴⁾ This tumor stained positively for anti-CD34 antibodies and factor VIII, but not for D2-40, which specifically recognizes lymphatic endothelial cells; therefore, lymphangioma was ruled out, and hemangioma was diagnosed.

Unfortunately, contrast imaging could not be performed because of bronchial asthma. MRI was not performed because we did not consider possibilities other

than a neurogenic tumor, which was also regretful. The risk of hemorrhage can be markedly reduced if mediastinal hemangioma can be predicted by imaging before surgery.

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