—Case Reports—

Venous Hemangioma of the Anterior Mediastinum

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Abstract

We report a rare case of venous hemangioma (VH) of the anterior mediastinum in a 56-year-old man admitted to our hospital because of hematemesis. Systemic examinations were performed and chest computer tomography (CT) revealed a 1.5-cm sized small nodule with contrast enhancement in the thymus. Both CT and magnetic resonance imaging (MRI) suggested a solid tumor such as a thymoma or neurogenic tumor rather than a vascular neoplasm. A partial thymectomy including this nodule by video-assisted thoracic surgery (VATS) was performed. Histological examination showed VH. There was no recurrence with no further treatment.
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Key words: mediastinum, venous hemangioma (VH), video-assisted thoracic surgery

Introduction

Venous hemangioma (VH) of the mediastinum is rare, accounting for 0.5% or less of all mediastinal tumors[1]. In our case, a VH was adjacent to the thymus and was suspected to be a solid tumor rather than a vascular neoplasm before surgery. We report of a case of VH in the anterior mediastinum and a review of the literatures concerning with the clinical and pathological features of this disease.

Case

A 56-year-old man was admitted because of hematemesis. Systemic examination revealed acute gastric mucosal lesion. There was no antecedent history of trauma. His past history was subarachnoidal hemorrhage (SAH) and clipping operation was performed five years ago. Laboratory findings were within normal limits. In these

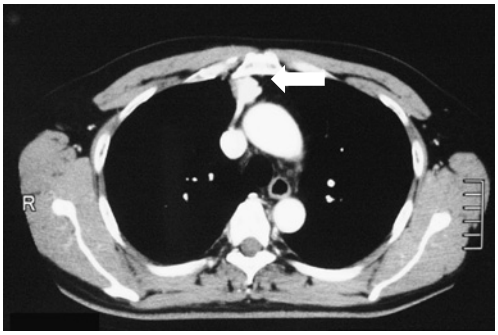

Fig. 1 Chest CT demonstrates a well-defined 1.5-cm nodule (**white arrow**) with moderate contrast. No calcification or fatty component can not be seen.

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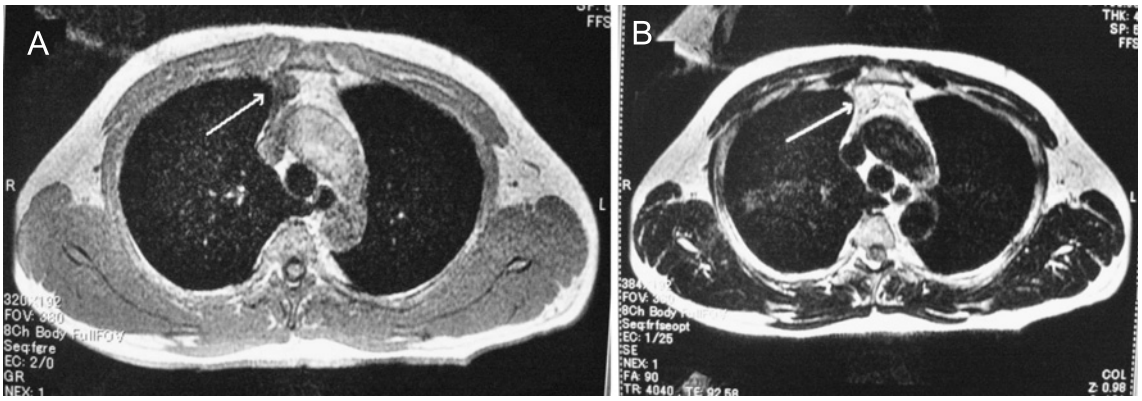

Fig. 2 MRI shows an isointense tumor with muscle on a T1-weighted image (**white arrow**, A) and high intensity on a T2-weighted image (**white arrow**, B).

examinations, chest CT had revealed a well defined 1.5-cm sized nodule with moderate contrast enhancement in the anterior mediastinum **(Fig. 1)**. MRI showed an isointense tumor with muscle on the T1-weighted image and high intensity on the T2-weighted image **(Fig. 2A, B)**. No calcification or fatty component was found in the lesion. The lesion was suspected to be a thymoma or neurogenic tumor rather than vascular neoplasm before surgery. Angiography could not be carried out by the refusal of patient. Systemic CT revealed no vascular disease in the brain, lung, liver, and other organs.

Both CT and MRI showed that the lesion was adjacent to the thymus. Our policy for possibly benign lesion is complete surgical resection with a part of thymus via video-assisted thoracic surgery (VATS) procedure rather than the median sternotomy. A 1-cm incision was respectively made in the third, fourth intercostal space, and fifth intercostal spaces to facilitate VATS. After applying a hemocrip to a small branch from right internal thoracic artery (RITA), we carefully dissected the lesion away from the superior vena cava (SVC). The specimen was removed with adequate margins including thymus tissues of at least 0.5 cm in all directions. Macroscopically, tumor presented a vascular lesion. The operating time was 165 minutes, and the intraoperative blood loss was 320 mL.

The resected tumor measured 1.5 × 1.0 cm and a cross section showed a vascular-like structure and formation of thrombus **(Fig. 3)**. Histological examination showed an enlarged lumen, lined with

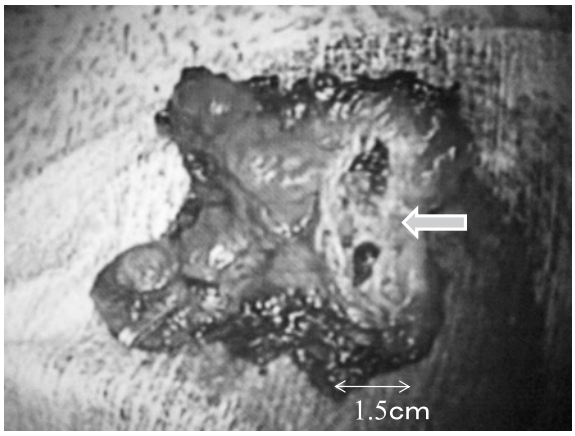

Fig. 3 Macroscopic appearance of a cut surface: the cystic space is surrounded by connective tissue and coagulative material can be seen in the cystic space.

smooth muscle cells in the adipose tissue; the cyst wall was composed of a large vein-type structure with thrombotic material **(Fig. 4A, B)**, and a transitional area was observed between the apparent large, vein-type wall and a thin-walled vessel **(Fig. 4C)**. On the basis of these histological findings, we diagnosed VH.

The postoperative course was uneventful, and the chest tube was removed on postoperative day (POD) 1. The patient was discharged with no complications on POD 5 and during 15 months of follow-up no recurrence was detected.

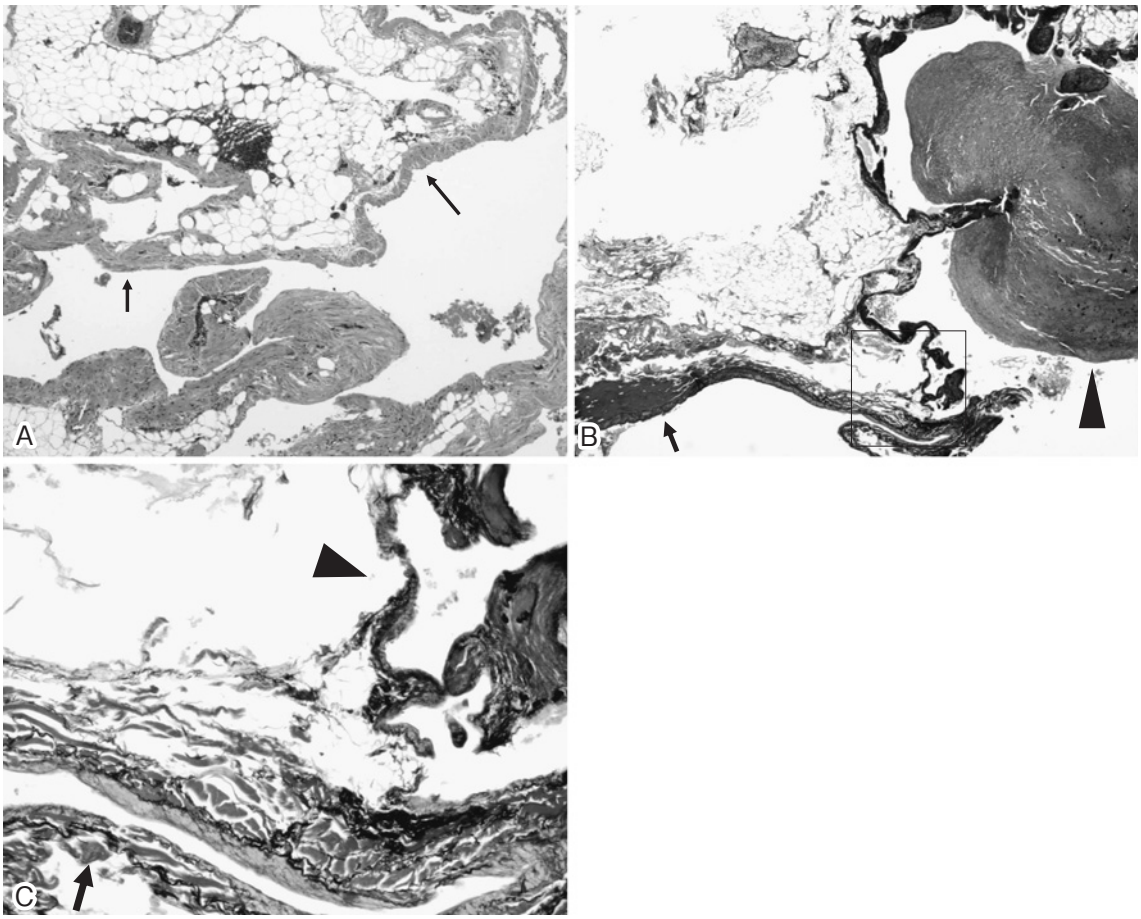

Fig. 4 Microscopic findings

A: The cystic space is lined with a thin layer of smooth muscle cells **(arrow)**. (Hematoxylin and eosin stain)

B: The wall of the cyst consists of a large vein-type structure. A vena cava-like structure **(arrow)** and thrombotic material **(arrowhead)** can be observed. (Elastica Van Gieson stain)

C: High magnification view of the square area in the **B**: a transitional area between the apparent large vein-type wall **(arrow)** and a thin-walled vessel **(arrowhead)** is observed. (Elastica Van Gieson stain)

Discussion

Mediastinal hemangiomas, which accounts for approximately 0.5% of all mediastinal tumors, are almost benign[1]. About 90% of benign blood vascular neoplasms in the mediastinum are capillary or cavernous hemangiomas[2]. In a nationwide survey of 1,546 mediastinal tumors, Wada et al. found that only 5 (0.32%) were hemangiomas [2]. Vascular malformations of the mediastinum include VHs, arteriovenous hemangiomas (AVM), hemangiomas, angiolipomas, angio-fibromas, glomus tumors, and hemangiopericytomas[2]. These lesions may provoke symptoms when they are infectious or when they exert pressure on neighboring structures after becoming enlarged; they can be detected incidentally on a chest radiographs or chest CTs[3,4]. In our case the patient had no symptoms and the VH was found during the evaluation of hematemesis.

Hemangiomas are usually round and lobulated with smooth margins and are often associated with phleboliths on X-ray imaging[5]. Additionally, on CT scanning, hemangiomas may show a blood flow and vascular channels or homogenous low-attenuation masses and cavernous hemangiomas may show characteristic "puddles" of enhancement following administration of a contrast agent[6]. According to McAdams et al., a characteristic of mediastinal hemangioma is that the center of the tumor shows stronger enhancement than the margin on CT[7], but we could not observe this finding in our case.

Generally, a preoperative diagnosis of this disease is difficult, and a correct diagnosis was not done in any of the 15 cases except two Japanese cases[1]. In our case, the lesion was a lobulated and well-defined nodule. On imaging diagnosis we suspected a solid tumors rather than a vascular neoplasm, however intraoperative macroscopic examination clearly showed a vascular lesion with thrombus. The formation of thrombus in a tumor might cause the misleading findings on imaging.

Differential diagnosis of the lesion includes other vascular mediastinal tumors such as cavernous hemangioma and angiolipoma, which are found in children and they appear as well-defined round or lobulated masses[8,9]. The VH in our patient could easily be differentiated from a cavernous hemangioma which also has large dilated vascular spaces. Angiolipoma is a rare benign tumor composed of mature adipose tissue and blood vessels; it is usually located in the subcutaneous tissue but can also be found occasionally in other locations[9].

Our treatment strategy was surgery rather than observation, because thymoma could not be ruled out. As the lesion was adjacent to the thymus, we considered partial thymectomy necessary to extirpate the lesion completely. After consideration of the lesion location, the extirpation by VATS might be a good option to treat this condition. Pathologically, the lesion was completely removed and the amount of intraoperative bleeding was acceptable.

In conclusion, we successfully removed a VH in the anterior mediastinum by means of VATS. Preoperative diagnosis of mediastinal hemangioma is difficult with imaging studies, however the possibility of hemangioma should be considered in patient with mediastinal tumors.

References

1. Cohen AJ, Sbaschnig RJ, Hochholzer L, et al.: Mediastinal hemangiomas. Ann Thorac Surg 1987; 43: 656–659.
2. Wada H, Teramatsu T: Mediastinal tumors-a statistical nationwide report of 1,546 cases between July, 1975 and May, 1979 in Japan. Nippon Kyobu Geka Gakkai Zasshi 1982; 30: 374–378.
3. Lunde P, Sorlie D, Botz KD, et al.: Huge arteriovenous malformation in the mediastinum. Clinical presentation and surgical management. Scand J Thoracic Cardiovasc Surg 1984; 18: 75–80.
4. Cheng SL, Wang HC, Yang PC: Posterior mediastinal arteriovenous malformation with atypical color Doppler sonographic findings. J Clin Ultrasound 2003; 31: 44–47.
5. Davis JM, Mark GJ, Greene R: Benign blood vascular tumors of the mediastinum: Report of four cases and review of the literature. Radiology 1978; 126: 581.
6. Papagiannopoulos K, Sheppard MN, Goldstraw P: Thymic hemangioma presenting with recurrent pleural effusion. Ann Thorac Surg 2000; 70: 297–299.
7. McAdams HP, Rosado-de-Christenson ML, Moran CA: Mediastinal hemangioma: Radiographic and CT features in 14 patients. Radiology 1994; 193: 399–402.
8. Lee SY, Lee Jh, Hur GY, et al.: Successful removal of a slowly growing mediastinal cavernous haemangioma after vascular embolization. Respirology 2006; 11: 493–495.
9. Sabharwal Gk, Strouse PJ: Posterior mediastinal hemangioma. Pediatr Radiol 2005; 35: 1263–1266.
10. Kline ME, Patel BU, Agosti SJ: Noninfiltrating angiolipoma of the mediastinum. Radiology 1990; 175: 737–738.

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