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Case Report

Giant esophageal hemangioma diagnosed by ^{99m}Tc-HSA-D scintigraphy following equivocal CT, MRI, and endoscopy[☆]

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ABSTRACT

Mediastinal cavernous hemangioma is a rare lesion requiring diagnosis without invasive procedure due to the risk of hemorrhage, which can be massive and even fatal. Here we describe the successful diagnosis of such a lesion using technetium-99m diethylenetriamine penta-acetic acid human serum albumin (^{99m}Tc-HSA-D) scintigraphy. A 36-year-old female with a 3-week back pain underwent endoscopic ultrasonography, contrast-enhanced CT, and MRI dynamic study which together revealed a submucosal tumor of the esophagus; likely to be either hemangioma or lymphangioma. Because of poor or no enhancement, it was impossible to distinguish the nature of the lesion. However, using delayed blood-pool imaging of ^{99m}Tc-HSA-D (at 40 minutes postinjection), and the characteristic accumulation, the tumor was clearly identifiable as an esophageal hemangioma. This case shows ^{99m}Tc-HSA-D scintigraphy to be an effective noninvasive imaging method to capture the characteristic hemodynamics of hemangioma.

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Introduction

Mediastinal cavernous hemangioma is a rare lesion caused by congenital vascular dysplasia. It accounts for less than 0.5% of mediastinal tumors [1]. Although these lesions may occur at any level within the esophagus, the lower esophagus is

the most common. Generally, most patients with esophageal hemangiomas have no clinical symptoms or signs; however, the symptoms of obstruction and hemorrhage, eg, dysphagia, dyspnea, hematemesis, and melena may occur depending on the tumor size or location [2]. Hemorrhage from the tumor can be massive and even fatal, therefore making the diagnosis using a noninvasive procedure, such as ^{99m}Tc-HSA-D

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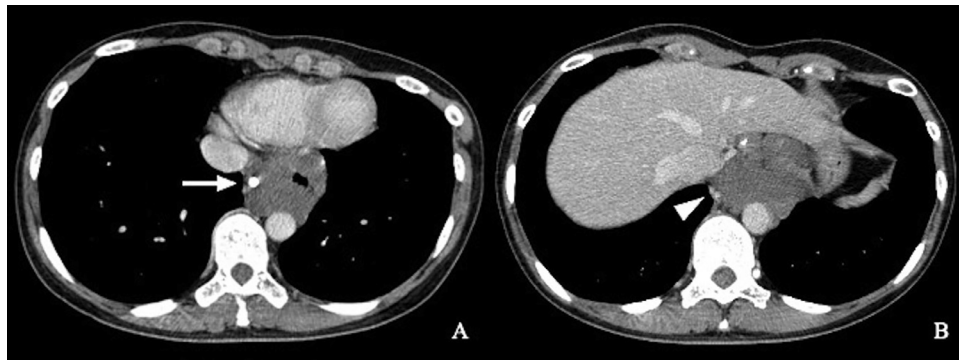


Fig. 1 – CECT was performed at 90 sec postinjection. Tumor was located at the dorsal side of the heart (A) and the left lobe of the liver (B). There were some nodular calcifications in the mass indicating phleboliths (arrows) of the hemangioma and peripheral puddling (arrowhead) of contrast medium, but the majority of the tumor was not enhanced.

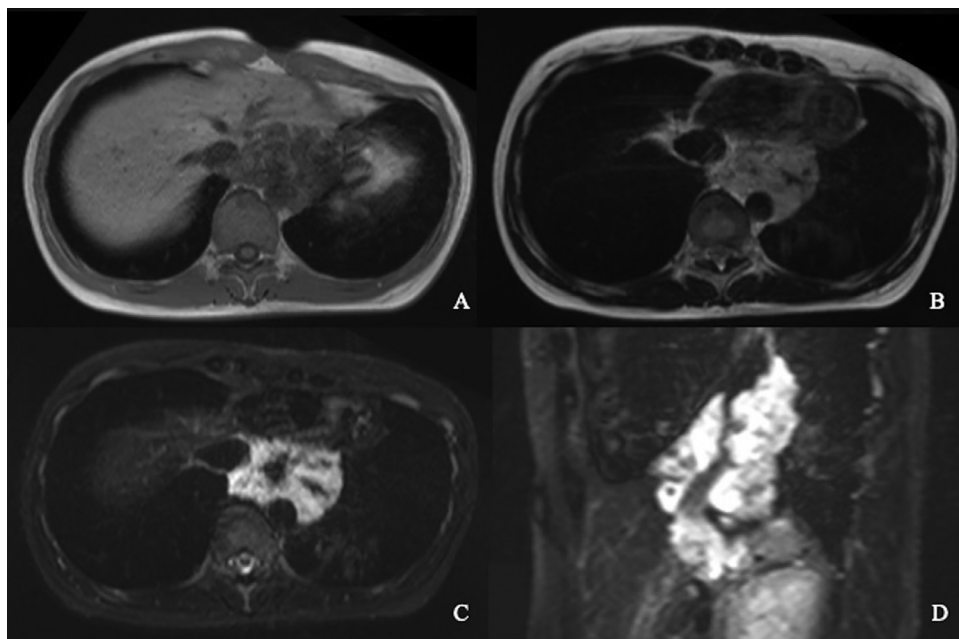


Fig. 2 – MRI axial T1-weighted image (A), axial T2-weighted image (B), and axial (C) and sagittal (D) STIR images. Multilocular lesion existed surrounding the esophagus. Markedly high intensity at the lesions by STIR was consistent with long T1 and T2 relaxation time showing the lesion is fluid-filled.

scintigraphy, is critical. After diagnosis, there are several treatment options available including esophagectomy, endoscopic removal, sclerotherapy, and laser therapy [2]. Of these methods, the approach deemed the least invasive, as judged on a case specific basis, is the preferred treatment. Here, we report a case of a mediastinal lesion which was diagnosed a giant esophageal hemangioma using ^{99m}Tc -HSA-D scintigraphy.

Case report

A 36-year-old female with a 3-week back pain was suspected of having a mediastinal lesion on plain CT of the thorax. She

had no medical history. Her family history and blood biochemistry test results were unremarkable.

Contrast-enhanced CT (CECT) performed at 90 seconds postinjection showed a poorly enhanced or nonenhanced mediastinal soft tissue mass, approximately $4.3 \times 6.6 \times 10.7$ cm, with an irregular margin along the thoracic and the abdominal esophagus, from the level of the carina to the cardiac junction. While there were some nodular calcifications and peripheral puddling of contrast medium, the majority of the tumor was not enhanced on the CECT image (Fig. 1A-B).

MRI showed inhomogeneous low intensity on T1-weighted image and relatively high intensity on T2-weighted image. Markedly high intensity at the lesions in STIR images was consistent with long T1 and T2 relaxation times, indicating the lesion was fluid-filled (Fig. 2A-D).

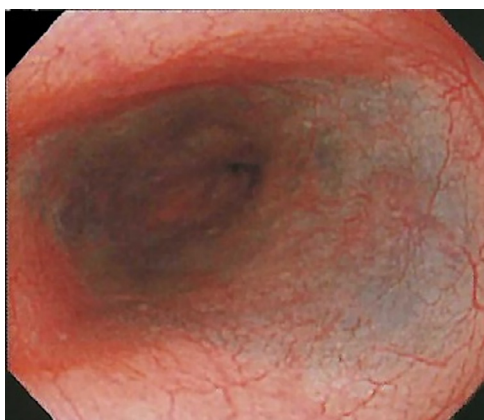


Fig. 3 – Endoscopy of the lower thoracic esophagus showed a blue color lesion distributed throughout an almost normal mucosa.

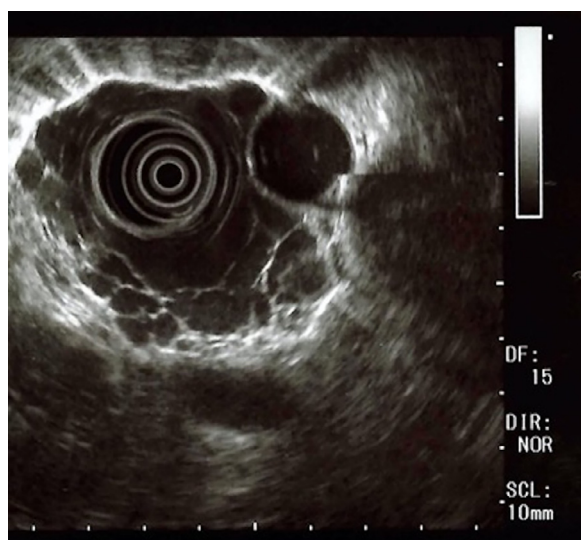


Fig. 4 – EUS showed homogenous anechoic or hypoechoic multilocular lesions in the submucosa layer.

Endoscopy of the lower thoracic esophagus showed a blue color lesion distributed throughout mucosa with a normal surface pattern, indicating blood-containing submucosal tissue (Fig. 3). There was no epithelial lesion. Endoscopic ultrasonography (EUS) showed homogenous anechoic or hypoechoic multilocular lesions with a smooth margin, neither nodular nor serrated, around the submucosa layer (Fig. 4). These findings were suggestive of a benign multilocular lesion, such as hemangioma or lymphangioma, arising from layers deeper than those of the mucosa.

To determine the tumors identity, a ^{99m}Tc -HSA-D dynamic study was performed until 40 minutes after administration followed by continuous delayed blood-pool SPECT/CT imaging. Radiotracer accumulation gradually increased just

below the cardiac pool in the middle of the abdomen (Fig. 5). SPECT/CT performed 40 min after administration revealed diffuse accumulation to the mass around the esophagus (Fig. 6), which suggested that the tumor was hemangioma and not lymphangioma. Finally, this tumor was found to be a circumferential tumor around the esophagus which was large along the cephalic-caudal axis and was extremely rich in blood pooling. Since the tumor was confirmed to be either cavernous or venous hemangioma, it was determined that thoracotomy was needed.

Discussion

Histologically, hemangiomas are classified into cavernous type, capillary type, and mixed type. They have been categorized as low-flow venous malformations since the International Society for the Study of Vascular Anomalies (ISSVA) in 2014 [3,4].

Decreased perfusion in a flow study and increased local blood pool activity in a late phase study by ^{99m}Tc -HSA-D scintigraphy is a feature often referred to as “perfusion/blood-pool mismatch”, which is one of the characteristic findings of cavernous type. Persistently increased uptake on a delayed scan is also a common finding. On the other hand, a capillary-type hemangioma is thought to show increased activity on both the perfusion image and blood-pool image because of its high blood flow [5].

In enhanced CT scan, arterial phase, venous phase, and delayed phase can be acquired, however, it is hard to image them at appropriate timing and the imaging session is accompanied by repetitive exposure to radiation [6,7,8]. This time, the hemodynamics of the lesion was observed up to 40 minutes continuously with ^{99m}Tc -HSA-D. Planar image taken from the back showed the accumulation, which increased very slowly over time in the part overlapping the left lobe of the liver, although the upper half of the tumor is hidden by the high accumulation in the heart. Moreover, ^{99m}Tc -HSA-D SPECT/CT added the anatomical information to a blood-pool image. It revealed diffuse accumulation all around the esophagus from the level of the carina to the level of the esophagus cardiac junction.

^{99m}Tc -HSA-D is a stable blood-pool scanning agent with stability conferred through the interposition of diethylenetriamine penta-acetic acid (DTPA), a chelating agent, between HSA and ^{99m}Tc . Available for clinical use in Japan, ^{99m}Tc -HSA-D is nontoxic to the kidneys and therefore safe to use in patients with poor renal function. ^{99m}Tc -HSA-D scintigraphy may be an alternative to 3-phase CT for the regular surveillance of the hemodynamic analysis, such as differentiation of hemangiomas, slow-filling endoleaks, small-intestine hemorrhages, and protein-losing gastroenteropathy that 3-phase CT does not capture [9].

In conclusion, we captured the characteristic hemodynamics of hemangioma using ^{99m}Tc -HSA-D scintigraphy. Such a noninvasive diagnostic procedure is very important to avoid hemorrhage from the angiogenic tumor.

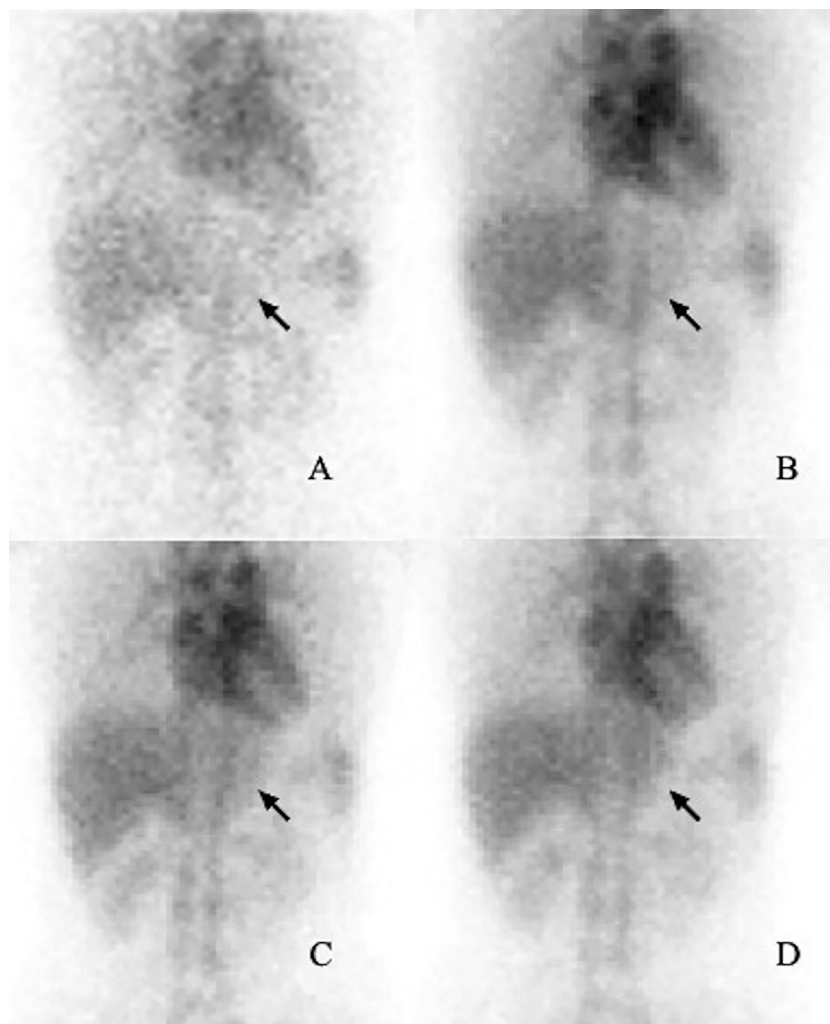


Fig. 5 – ^{99m}Tc -HSA-D dynamic study at 30 sec (A), 90 sec (B), 15 min (C), and 90 min (D) postinjection of ^{99m}Tc -HSA-D. Radiotracer accumulation in the tumor at the level of the left lobe of the liver was gradually increasing (arrow) although the accumulation in the right lobe of the liver showed little change. Accumulation in the intrathoracic lesion cannot be evaluated because of the overlapped cardiac blood pool.

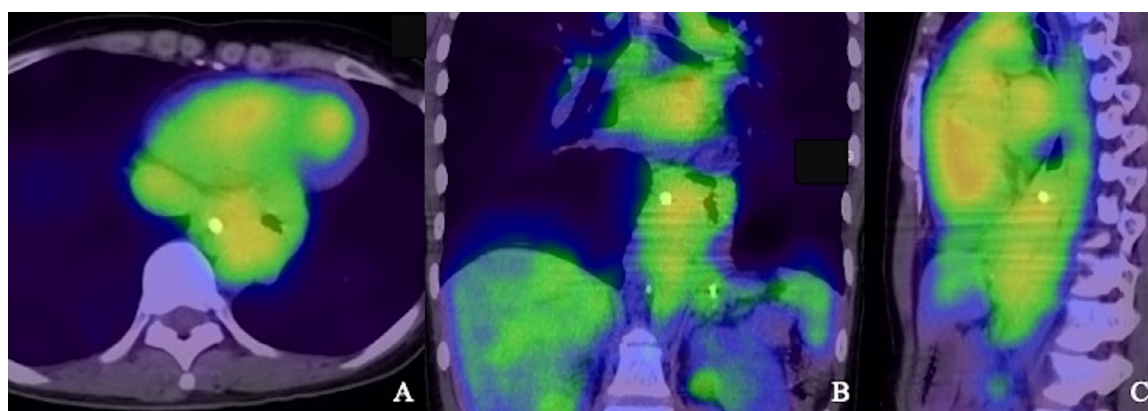


Fig. 6 – Axial (A), coronal (B), and sagittal (C) SPECT/CT image at 40 minutes postinjection of ^{99m}Tc -HSA-D. The diffuse accumulation in the tumor, which is almost the same extent as the cardiac accumulation, is shown.

Patient consent statement

It does not require patient consent for the case with no identifiable personal information.

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