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Case Report

Esophageal lymphangioma: Endoscopic ultrasound, computed tomography, and magnetic resonance imaging appearance ☆

Katsuya Nanjo, MD[a], *Daisuke Tsurumaru, MD, PhD*[b,*], *Masakazu Hirakawa, MD, PhD*[a], *Yusuke Nishimuta, MD*[b], *Koshi Mimori, MD, PhD*[c], *Kousei Ishigami, MD, PhD*[b]

[a] *Department of Radiology, Kyushu University Beppu Hospital, Beppu-city, Oita, Japan*
[b] *Department of Clinical Radiology, Graduate School of Medical Sciences, Kyushu University, Fukuokas-city, Fukuoka, Japan*
[c] *Department of Surgery, Kyushu University Beppu Hospital, Beppu-city, Oita, Japan*

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ABSTRACT

A case of esophageal lymphangioma in a 75-year-old man who complained of worsening dysphagia is presented. Endoscopic ultrasound showed an echogenic pattern of honeycomb or grid-like multiple microcysts within the submucosa. The sagittal image of computed tomography showed a thickened esophageal wall and fluid retention in the proximal esophageal lumen. Magnetic resonance imaging showed a high signal intensity mass with a septate-like internal structure on T2-weighted imaging and short tau inversion recovery. The tumor was completely resected by endoscopic submucosal dissection. Esophageal lymphangioma is a rare submucosal tumor that can be precisely diagnosed by CT and/or MRI.

Introduction

Lymphangiomas occur in various parts of the body, most commonly in the head and neck region, but rarely in the gastrointestinal tract, and they are especially rare in the esophagus [1]. There have been a few reports of the endoscopic ultrasound (EUS) findings of esophageal lymphangioma [2–4]. However, there seem to be no reports of its computed tomography (CT) and magnetic resonance imaging (MRI) features. Thus, a case of esophageal lymphangioma evaluated by EUS, CT, and MRI that was finally removed by endoscopic submucosal resection (ESD) is described.

Case report

A 75-year-old man who complained of dysphagia and nausea visited our hospital. He underwent upper endoscopy, which

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* Corresponding author.
E-mail address: tsurumaru.daisuke.931@m.kyushu-u.ac.jp (D. Tsurumaru).

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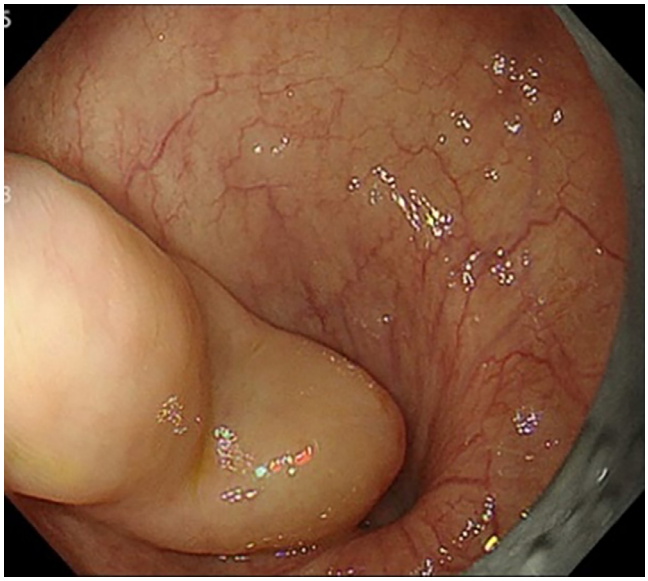

Fig. 1 – Upper endoscopy shows a polypoid submucosal tumor located in the lower esophagus.

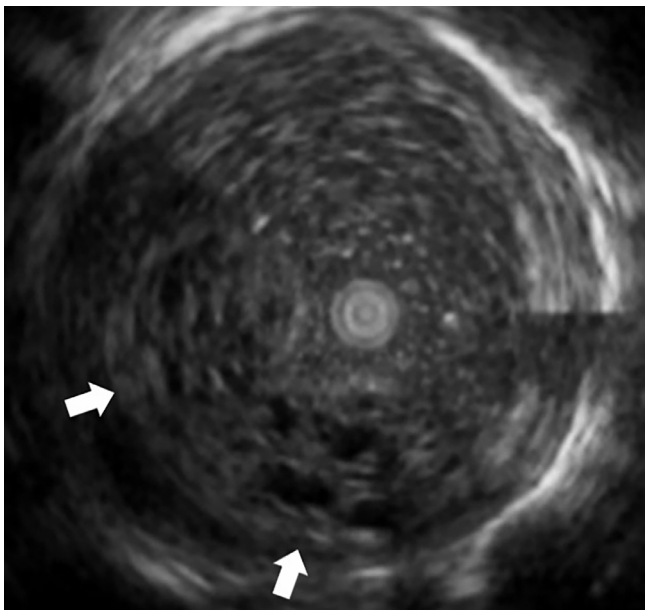

Fig. 2 – EUS shows an echogenic mass of honeycomb or grid-like multiple microcysts within the submucosa with no invasion to muscularis propria (arrows).

showed a submucosal tumor (SMT) estimated to be 4 cm in diameter in the lower esophagus (Fig. 1). The overlying mucosal surface was normal, without ulcer or erosion. Magnified narrow-band imaging showed no irregular changes of intrapapillary capillary loops. EUS showed an echogenic pattern of honeycomb or grid-like multiple microcysts within the submucosa, with no invasion to the muscularis propria (Fig. 2). Plain and enhanced CT showed low-attenuation tumor with no enhancement. The CT sagittal image showed a thickened esophageal wall and fluid retention in the proximal esophageal lumen, which suggested that the tumor had caused esophageal obstruction for a long period (Fig. 3). MRI showed a high signal intensity mass with septate-like internal structure on T2-weighted imaging and short tau inversion recovery (STIR). Diffusion-weighted imaging (DWI) and the apparent diffusion coefficient (ADC) map showed no diffusion restriction, which suggested less malignant behavior (Fig. 4). The patient was observed despite further diagnostic examinations to confirm pathology. Within a year and a half of observation, however, the patient returned because of worsening dysphagia. Noninvasive endoscopic treatment was performed because of the benign nature of the tumor. The tumor was completely resected by endoscopic submucosal dissection (ESD).

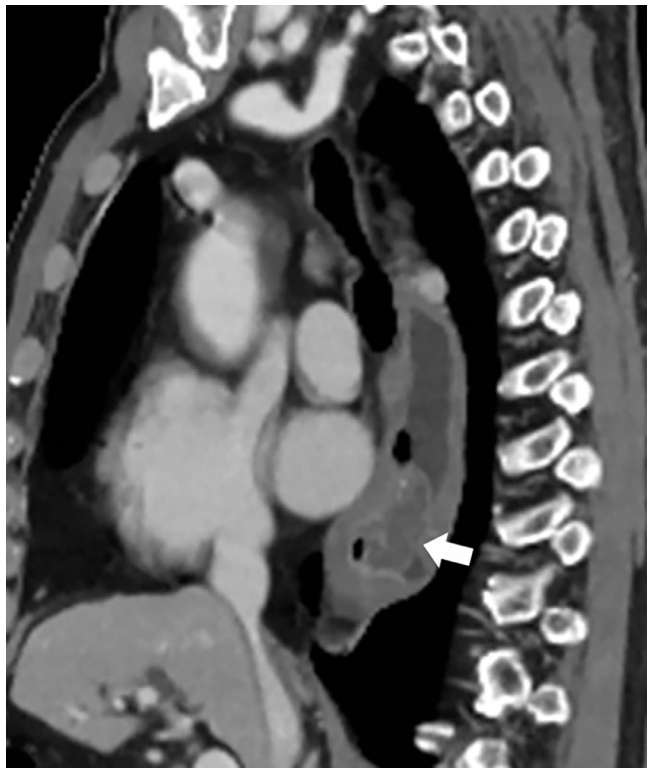

Fig. 3 – Contrast-enhanced CT (sagittal image) shows the tumor located in the lower thoracic esophagus (arrow). The esophageal wall is diffusely thickened, and fluid has collected in the proximal esophageal lumen.

Macroscopically, the resected specimen contained a whitish nodular lesion located mainly in the submucosa, 51×23 mm^2 in size. Microscopically, the tumor was composed of dilated lymphatic vessels of various sizes. The esophageal squamous epithelium showed no pathological change. On immunohistochemical staining, the lining of endothelial cells was positive for CD34 and D2-40. This feature confirmed the final diagnosis as lymphangioma of the esophagus. There were no signs of malignancy (Fig. 5). The patient's post-ESD course was uneventful. His dysphagia disappeared completely and did not recur within the 6-month follow-up period.

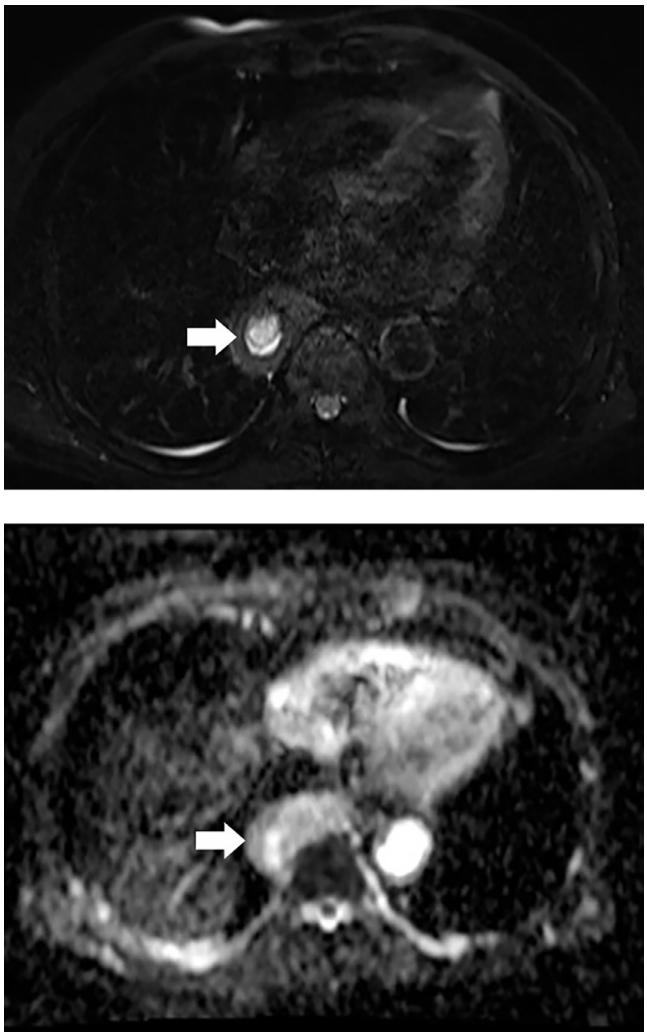

Fig. 4 – (A) MRI shows a high signal intensity mass with septate-like internal structure on STIR (arrow). (B) The ADC map shows no diffusion restriction (arrow).

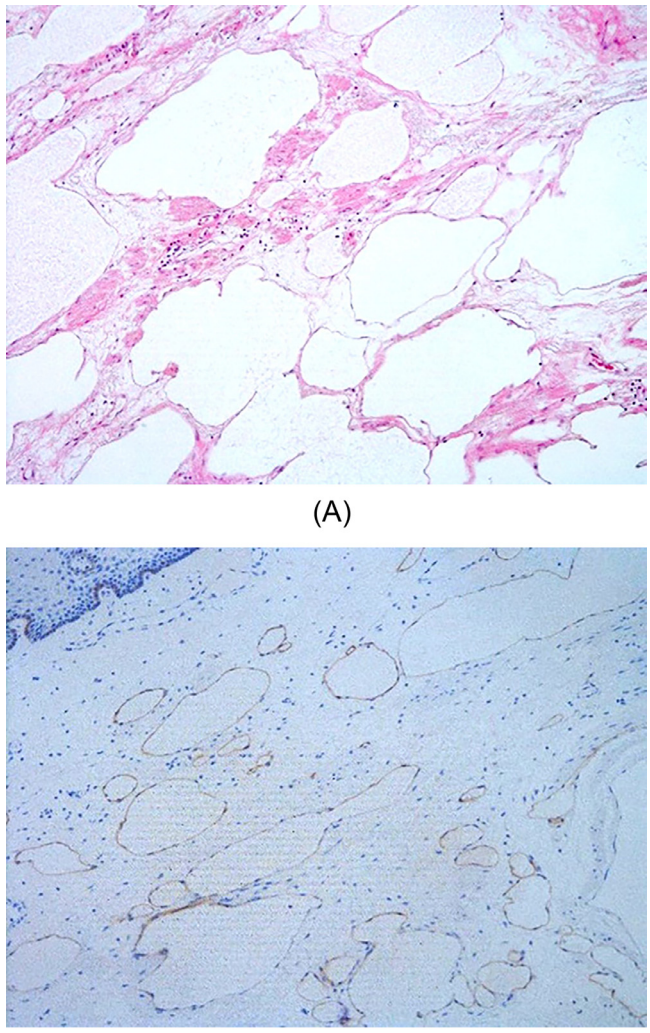

Fig. 5 – (A) Microscopy (hematoxylin and eosin, magnification ×200) shows tumor composed of dilated lymphatic vessels in various sizes underneath the normal esophageal squamous epithelium. (B) Endothelial cells are immunohistochemically positive for D2-40.

Discussion

Benign esophageal SMTs are rare and often discovered incidentally by screening endoscopy. Leiomyoma is the most common, followed by granular cell tumors, neurogenic tumors, and duplication cysts [5,6]. Endoscopy is not frequently diagnostic because invasive procedures such as EUS-guided fine-needle aspiration biopsy are needed to confirm pathology.

Esophageal lymphangioma is extremely rare. About 1% of lymphangiomas were originated in the gastrointestinal tract, of which the most frequent location was the colon, followed by the stomach, duodenum, small intestine, and esophagus [1]. There have been several case reports. EUS is a common diagnostic tool for esophageal lymphangioma, which shows an echogenic pattern of honeycomb or grid-like multiple microcysts, as in the present case [1]. EUS is also useful in differentiation from solid tumors such as gastrointestinal stromal tumors or leiomyomas. CT or MRI may be chosen to evaluate large tumors, as for the present case, although there have been no reports showing CT or MRI findings of esophageal lymphangioma. In the present case, CT was useful for evaluating the tumor component, surrounding tissue of the tumor, and complications related to obstruction using sagittal images. MRI was also useful for evaluating malignant behavior using DWI and ADC mapping. Imaging including EUS, CT, and MRI of esophageal duplication cysts are similar to esophageal lymphangioma. Duplication cysts are pure cystic lesion, however, lymphangiomas have honeycomb or grid-like multiple microcysts. These features might be differentiation point [6].

Clinical presentations of patients with esophageal lymphangioma are nonspecific. They may be asymptomatic or may have various complaints depending on the location and size of the tumor. As in the present case, dysphagia is the most common symptom. Other symptoms reported include heartburn, epigastric pain, and vomiting. Treatment options also

depend on the clinical presentation. Currently, with advances in endoscopic technology, noninvasive endoscopic treatments are recommended due to its benign etiology. ESD is suitable to completely resect a large tumor that could cause obstructive symptoms, as in the present case [7,8].

In conclusion, esophageal lymphangioma is a rare submucosal tumor that might cause symptoms depending on its location and size. Complications related to esophageal obstruction can be accurately evaluated by additional CT or MRI examinations.

Patient consent

Written informed consent for the publication of this case report was obtained from the patient.

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