Endoscopic submucosal dissection of a giant esophageal lymphangioma

Lymphangioma of the esophagus is an exceedingly rare submucosal benign tumor [1]. Only 23 cases have been reported since 1934, and most of the tumors are smaller than 2 cm in diameter. Herein, we present the first report of a giant esophageal lymphangioma (16 × 6 × 4 cm), which was completely and safely resected by endoscopic submucosal dissection (ESD).

A 46-year-old man suffering dysphagia for 3 years visited our department. Computed tomography revealed an irregular, intraluminal, and low-density mass (16 × 6 × 4 cm) in the middle-to-lower part of the esophagus (▶ Fig. 1c, d). Esophagogastroduodenoscopy revealed a giant, yellowish, and translucent submucosal lesion, with a lustrous surface and vesicular change in the esophagus at 28 cm from the incisor and extending into the lesser curvature of the cardia (▶ Fig. 1a). Endoscopic ultrasonography showed a grid-like hypoechoic structure with a heterogeneous echo that originated from the submucosal layer; the muscularis propria was intact. (c, d) Contrast-enhanced chest computed tomography revealed an irregular, intraluminal, and low-density mass (16 × 6 × 4 cm) in the middle-to-lower part of the esophagus.

ESD was performed to relieve the patient’s symptoms (▶ Video 1). First, circumferential marking was made using a dual knife (▶ Fig. 2a). Then, a solution of indigo carmine and glycerol was injected to lift the submucosa (▶ Fig. 2b). After circumferential mucosal incision, a clip with dental floss was placed directly onto the dissected submucosal tissue, which provided proper countertraction for a clear submucosal dissection plane during submucosal dissection (▶ Fig. 2c, d, e, g). The tumor was completely and uneventfully resected en bloc; however, because of its large size, we could not retrieve it en bloc, even under the vigorous support of specialist doctors. Given the benign nature of the tumor, it was finally divided into pieces in the esophagus and subsequently retrieved using a snare (▶ Fig. 2f, ▶ Fig. 3).

Histologically, the tumor showed irregularly dilated lymphatic vessels beneath squamous epithelium and the submucosa, with lymphoid fluid in the lumen (▶ Fig. 4a, b), and accompanying CD31 and D2-40 expression (▶ Fig. 4c, d). An esophageal lymphangioma was diagnosed.

At 2 weeks after surgery, triamcinolone acetonide (Kenacort, 40 mg/mL; Zhejiang Xianju Pharmaceutical Co., Ltd., Zhejiang, China), diluted 1:9 with saline, was injected at several points in the submucosa to prevent esophageal stenosis.
No residual tumor or stenosis was observed endoscopically at 4 months after treatment (Fig. 2 h, i).

Surgical resection is usually recommended for symptomatic esophageal lymphangioma larger than 2 cm [2]. To the best of our knowledge, this is the largest esophageal lymphangioma that has been completely and successfully removed using ESD.

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Competing interests
None

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Fig. 3 Macroscopic appearance of the tumor.

Fig. 4 Histology of the resected specimen showed irregularly dilated lymphatic vessels beneath squamous epithelium and the submucosa, with lymphoid fluid in the lumen. Hematoxylin and eosin: a × 40 magnification; b × 100 magnification. c Lymphatic epithelial cells were stained with CD31 antibody (× 100). d The resected mass was positive for D2-40 (× 100).

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