A 58-year-old man presented with an 8-year history of intermittent dysphagia. There was no other relevant medical history. A barium esophagogram revealed a stenotic lesion in the middle and lower thoracic esophagus with multiple intramural tracks [1]. Endoscopic examination revealed an annular stricture of the esophagus extending from 27 cm to 40 cm from the incisors, as well as multiple small orifices (Fig. 1).

A diagnosis of esophageal intramural pseudodiverticulosis (EIPD) was suspected. As the patient did not have diabetes, candidal esophagitis, or esophageal carcinoma, and because esophageal inflammation has been reported in up to 90% of patients with EIPD [2], we prescribed a proton-pump inhibitor to suppress gastric acid secretion. However, after 7 months, the patient could barely swallow food, and repeat endoscopy showed that the lesions had clearly progressed. The multiple diverticula had enlarged and the lumen was criss-crossed by multiple mucosal bridges (Fig. 2).

Computed tomography of the chest showed marked thickening of the esophageal wall with intramural air sacs running parallel to the lumen of the upper to lower thoracic esophagus (Fig. 3).

A subtotal esophagectomy was carried out in view of the recent worsening of symptoms and endoscopic findings. On histological examination, the esophageal wall was remarkably thickened by submucosal fibrosis and hypertrophic muscularis propria. Dilated excretory ducts were seen extending from the mucosal epithelium to the submucosal layer (Fig. 4).

There were no features of malignancy in the esophageal wall or the regional lymph nodes, and a final diagnosis of EIPD was made. After surgery, the patient’s dysphagia improved, and he has remained asymptomatic on follow up.

To our knowledge, this is the first report of EIPD with a hypertrophic muscular layer in the esophageal wall, as demonstrated in a full-thickness histological specimen [3, 4]. Furthermore, with serial endoscopy, we observed the progression of EIPD in the short term.
Fig. 4 Photomicrograph of the resected surgical specimen (hematoxylin and eosin, magnification × 40). The esophageal wall is markedly thickened by submucosal fibrosis and the hypertrophic muscularis propria. Dilated excretory ducts extend from the mucosal epithelium to the submucosal layer. A keratin component, inflammatory cells, and fungal hyphae are seen in, and lymphocytes and plasma cells around, the ducts.

References

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Corresponding author
M. Tajika, MD, PhD
Department of Endoscopy
Aichi Cancer Center Hospital
1-1 Kanokoden
Chikusa-ku
Nagoya 464-8681
Japan
Fax: +81-52-7642942
mtajika@aichi-cc.jp

J. Tsuboi1,2, M. Tajika3, T. Nakamura4, H. Kawai5, V. Bhatia4, R. Takayama1, J. Kitamura5, Y. Yatabe5, S. Hatooka6, M. Shinoda6, K. Yamao1, Y. Ogura2
1 Department of Gastroenterology, Aichi Cancer Center Hospital, Nagoya, Japan
2 Department of Gastroenterology, Matusaka City Hospital, Matusaka, Japan
3 Department of Endoscopy, Aichi Cancer Center Hospital, Nagoya, Japan
4 Department of Medical Hepatology, Institute of Liver and Biliary Sciences, New Delhi, India
5 Department of Pathology and Molecular Diagnosis, Aichi Cancer Center Hospital, Nagoya, Japan
6 Department of Thoracic Surgery, Aichi Cancer Center Hospital, Nagoya, Japan