

CASE REPORT

Endoscopic submucosal dissection for superficial esophageal cancer in a diverticulum: A case report with literature review

Yasuhiko Hamada, Yohei Ikenoyama, Yuhei Umeda, Hiroki Yukimoto, Akina Shigefuku, Yasuko Fujiwara, Tsuyoshi Beppu, Misaki Nakamura, Noriyuki Horiki and Hayato Nakagawa

Department of Gastroenterology and Hepatology, Mie University Hospital, Tsu, Japan

Key words

diverticulum, endoscopic submucosal dissection, endoscopic ultrasonography, esophageal cancer, traction.

Accepted for publication 3 December 2023.

Correspondence

Yasuhiko Hamada, Department of Gastroenterology and Hepatology, Mie University Hospital, 2-174 Edobashi, Tsu 514-8507 Japan. Email: y-hamada@med.mie-u.ac.jp

Declaration of conflict of interest: The authors declare no conflict of interest.

Author contribution: Yasuhiko Hamada contributed to the conception and drafting of the article. Yohei Ikenoyama, Yuhei Umeda, Hiroki Yukimoto, Akina Shigefuku, Yasuko Fujiwara, Tsuyoshi Beppu, Misaki Nakamura, Noriyuki Horiki, and Hayato Nakagawa contributed to critical revision of the article for important intellectual content. All authors approved the final draft of the manuscript.

Financial support: The authors did not receive any funding for the study relevant to this article.

Introduction

Superficial esophageal cancer (SEC) in a diverticulum is rare and has a high risk of perforation during endoscopic resection due to the lack of muscle layer.¹ Therefore, the standard treatment for this cancer is surgery or irradiation²; however, these treatments have a disadvantage of over-invasiveness. Currently, endoscopic submucosal dissection (ESD) is a standard treatment for SEC because of its minimal invasiveness and good effectiveness.³ ESD for SEC in a diverticulum is challenging for endoscopists, and the indication of ESD should be determined with caution.

This case report discusses a patient with SEC in a diverticulum treated by ESD and includes a review of the literature.

Case report

A 79-year-old male patient with a history of ESD for gastric cancer underwent surveillance esophagogastroduodenoscopy (EGD). He also had a history of prostate cancer. The patient's physical

Abstract

Superficial esophageal cancer (SEC) in a diverticulum is rare and has a high risk of perforation during endoscopic resection. Although endoscopic submucosal dissection (ESD) is a standard treatment option, it is challenging to perform. Here, we describe the case of a 79-year-old male patient with a history of ESD for SEC. Surveillance esophagogastroduodenoscopy identified a 20-mm-sized reddish depressed lesion in a diverticulum in the middle esophagus. The lesion was confirmed to be squamous cell carcinoma by biopsy. Magnification endoscopy with narrow-band imaging showed intraepithelial papillary capillary loops of type B1 according to the magnified endoscopic classification of the Japan Esophageal Society. Endoscopic ultrasonography revealed the presence of the muscular layer of the esophagus wall in the diverticulum. Therefore, the lesion was diagnosed as SEC, confined to the epithelium or lamina propria mucosae, in a Rokitansky diverticulum. Based on these findings, ESD was considered technically feasible. Traction-assisted ESD using clip with line was performed, and en bloc resection was achieved without adverse events. The resected specimen pathologically revealed a squamous cell carcinoma confined to the lamina propria mucosae without lymphovascular invasion, suggesting a curative resection. The patient recovered well, and no recurrence has been observed for 5 years after the ESD. Whether ESD is appropriate for the treatment of SEC in a diverticulum remains unclear. However, our case shows that it can be a treatment option in such cases due to its minimal invasiveness and good effectiveness.

examination and laboratory tests were unremarkable. The EGD identified a 20-mm-sized reddish depressed lesion in a diverticulum in the middle esophagus (Fig. S1a, Supporting information; arrows). Narrow-band imaging (NBI) endoscopy identified the lesion as a well-demarcated brownish area (Fig. S1b; arrows). The lesion was confirmed by biopsy to be squamous cell carcinoma. Magnification endoscopy with NBI showed intraepithelial papillary capillary loops of type B1 according to the magnified endoscopic classification of the Japan Esophageal Society (Fig. S1c).⁴ Endoscopic ultrasonography (EUS) did not reveal any lack of muscular layer of the esophagus wall in the diverticulum (Fig. S1d; arrows). Contrast-enhanced computed tomography (CT) revealed an esophageal diverticulum near the tracheal bifurcation (Fig. S2a, Supporting information, axial image; S2b, coronal image; arrow). The CT also revealed no lymph node swelling or distant metastasis. Based on these findings, we diagnosed the lesion as SEC, confined to the epithelium or lamina propria mucosae, in a Rokitansky diverticulum. ESD was

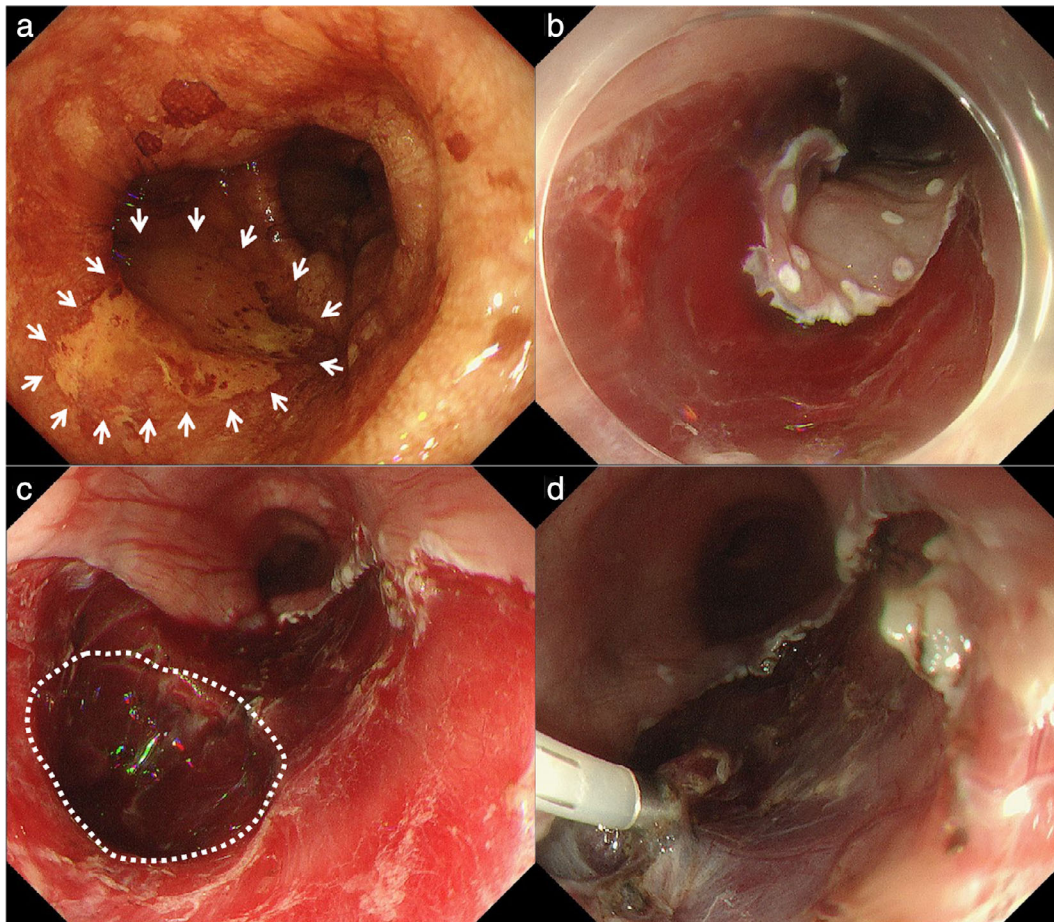


Figure 1 Endoscopic findings during endoscopic submucosal dissection (ESD). (a) Lugol chromoendoscopy identifying the lesion area (arrows). (b) Circumferential incision being performed. (c) A muscular defect (white dotted line) in the center of the post-ESD ulcer. (d) The muscular defect being completely closed endoscopically using endoclips.

considered technically feasible. After discussion with the patient, ESD was performed with the DualKnife (KD-655Q; Olympus Optical Co., Ltd., Tokyo, Japan) (Fig. 1a–d). We performed a submucosal injection of hyaluronic acid and an initial incision of the mucosa outside of the diverticulum to prevent perforation. The submucosal dissection was shallow, which further decreased this risk. After the circumferential incision, we anchored a clip with line to the oral edge of the resected mucosa. During dissection, a 10-mm-sized muscular defect appeared through the transparent submucosa, but the clip-with-line traction made it possible to dissect the submucosa over the muscular defect area without perforation. Finally, en bloc resection was achieved in 86 min without adverse events. The muscular defect was closed endoscopically using endoclips. Pathological examination revealed a squamous cell carcinoma confined to the lamina propria mucosae without lymphovascular invasion, suggesting a curative resection (Fig. S3a, Supporting information, resected specimen; Fig. S3b, pathological image). A soft diet was initiated on postoperative day 7, and the patient was discharged on postoperative day 11. Follow-up EGD revealed that the post-ESD ulcer had

healed completely, and no recurrence was noted for 5 years post ESD.

Discussion

Esophageal diverticula are rare with a prevalence of 0.06–3.6% based on radiologic and endoscopic analyses.^{2,5} Classically, the Zenker's and epiphrenic diverticula are pulsion diverticula and the Rokitansky diverticulum is a traction diverticulum. The incidence of cancer arising in the esophageal diverticulum is between 0.3% and 0.8% of all esophageal cancers.⁶ In general, the standard treatment for an SEC in a diverticulum is surgery or irradiation because the esophageal wall in the diverticulum sometimes lacks the muscle layer, and endoscopic resection has a higher risk of perforation.² However, the Rokitansky diverticulum usually includes all the normal layers of the esophagus,² and endoscopic resection can be used for SECs in such a diverticulum.

A MEDLINE search of the well-documented literature in English language up to 2023 using the search terms

“diverticulum” and “esophageal cancer” revealed 10 studies (including the current study) that included SEC in a diverticulum treated with ESD (Table S1, Supporting information; the reference articles are described in Doc. S1, Supporting information). The mean age at diagnosis was 68.3 years (range: 55–80 years). The main lesion location was the middle esophagus, followed by the lower esophagus. Among the six cases with well-documented lesion size, the mean diameter was 21.8 mm (range: 5–28 mm). The main lesion morphology was a depressed type (7/10, 70.0%). The invasion depth was confined to the mucosa in all cases. Muscular defect was seen in four (40.0%) cases. Among the five cases with well-documented endoknife use during the ESD procedure (Table S1), a needle-type endoknife was used in four cases (DualKnife in three cases, including ours, and FlushKnife BT [Fujifilm Medical Co., Ltd., Tokyo, Japan] in one case), whereas a scissor-type endoknife was used in one case (SB Knife [SB-Kawasumi, Inc., Kanagawa, Japan]). Emphysema was seen in three (30.0%) cases postoperatively; however, all the cases were managed conservatively.

To date, it is controversial whether ESD is appropriate for the treatment of SEC in a diverticulum; however, it can be a potential treatment option due to its minimal invasiveness and good effectiveness.⁷ In this case, we chose ESD as the treatment for the following reasons: (i) we have considerable experience (over 300 cases) of performing ESD for SECs, and closing the resulting perforated defect with hemoclips is usually feasible; (ii) use of a traction technique in esophageal ESD reduces the risk of perforation,⁸ and previous studies have reported that SEC in a diverticulum can be safely treated by traction-assisted ESD^{1,9,10}; and (iii) preoperative EUS showed the presence of the muscle layer of the esophagus wall inside the diverticulum; however, there was a possibility that complete mapping of the entire diverticulum had not been achieved.

When ESD is performed to treat SEC in a diverticulum, the risk of complications, such as perforation, emphysema, and pneumothorax, should be thoroughly explained preoperatively, and measures should be taken to prevent perforation. In this case, the traction technique was useful for visualizing the incision line. In addition, we performed submucosal injection of hyaluronic acid and an initial incision of the mucosa outside of the diverticulum to prevent perforation. The submucosal dissection was shallow, which further decreased the risk. When a muscular defect is detected in a diverticulum during ESD, it is desirable to close the defect with endoclips, if possible. Tsuji *et al.* reported the usefulness of placing polyglycolic acid sheets onto an ESD ulcer with a muscular defect in an esophageal diverticulum.¹ Understandably, it is also necessary to prepare supplies, such as a nasogastric tube and thoracic drain, in case of perforation, emphysema, and pneumothorax.

In conclusion, we reported a case of SEC in a diverticulum treated with ESD. The lesion was completely removed without adverse events, and no recurrence was observed for 5 years. Although the usefulness of ESD for the treatment of SEC in a diverticulum remains controversial because of the possibility of muscle layer defects, it can be considered in cases such as ours due to its minimal invasiveness and good effectiveness. Nevertheless, adequate preoperative evaluation and treatment planning are warranted.

Acknowledgment

We would like to thank Editage (www.editage.com) for English language editing.

Ethics approval statement

All procedures followed were performed in accordance with the ethical standards laid down in the Declaration of Helsinki and its later amendments.

Patient consent statement

Informed consent was obtained from the patient for publication of this case report and accompanying images.

Data availability statement. The data that support the findings of this article are available from the corresponding author upon reasonable request.

References

- 1 Tsuji Y, Saito I, Koike K. Traction-assisted esophageal endoscopic submucosal dissection for treatment of squamous cell carcinoma involving a diverticulum. *Dig. Endosc.* 2019; **31**: e7–8.
- 2 Herbella FA, Dubecz A, Patti MG. Esophageal diverticula and cancer. *Dis. Esophagus.* 2012; **25**: 153–8.
- 3 Kitagawa Y, Ishihara R, Ishikawa H *et al.* Esophageal cancer practice guidelines 2022 edited by the Japan esophageal society: part 1. *Esophagus.* 2023; **20**: 343–72.
- 4 Oyama T, Inoue H, Arima M *et al.* Prediction of the invasion depth of superficial squamous cell carcinoma based on microvessel morphology: magnifying endoscopic classification of the Japan Esophageal Society. *Esophagus.* 2017; **14**: 105–12.
- 5 Watanabe S, Matsuda K, Arima K *et al.* Detection of subclinical disorders of the hypopharynx and larynx by gastrointestinal endoscopy. *Endoscopy.* 1996; **28**: 295–8.
- 6 Fujita H, Kakegawa T, Shima S, Kumagaya Y. Carcinoma within a middle esophageal (parabronchial) diverticulum: a case report and the review of the literature. *Jpn. J. Surg.* 1980; **10**: 142–8.
- 7 Gotoda T, Takenaka R, Inokuchi T *et al.* Endoscopic submucosal dissection for a superficial esophageal squamous cell carcinoma within the parabronchial diverticula. *Dig. Endosc.* 2013; **25**: 91–2.
- 8 Yoshida M, Takizawa K, Nonaka S *et al.* Conventional versus traction-assisted endoscopic submucosal dissection for large esophageal cancers: a multicenter, randomized controlled trial (with video). *Gastrointest. Endosc.* 2020; **91**: 55–65.e2.
- 9 Esaki M, Suzuki S, Gotoda T. Endoscopic submucosal dissection of superficial esophageal cancer expanding into the diverticulum. *Dig. Endosc.* 2019; **31**: e30–1.
- 10 Tanaka K, Yabuuchi Y, Yamashita D, Hosotani K, Morita S, Inokuma T. Endoscopic submucosal dissection for a superficial esophageal squamous cell carcinoma located in a Rokitansky diverticulum. *Endoscopy.* 2022; **54**: E986–7.

Supporting information

Additional supporting information may be found in the online version of this article at the publisher's website:

Doc. S1. Reference articles for the cases described in Table S1.

Figure S1. Esophagogastroduodenoscopy findings. (a) Esophagogastroduodenoscopic identification of a 20-mm-sized reddish depressed lesion in a diverticulum in the middle esophagus (arrows). (b) Narrow-band imaging endoscopic identification of the lesion as a well-demarcated brownish area (arrows). (c) Magnification endoscopy with narrow-band imaging showing intraepithelial papillary capillary loops of type B1 according to the magnified endoscopic classification of the Japan Esophageal Society. (d) Endoscopic ultrasonography revealing the presence of the muscle layer (arrows) of the esophagus wall in the diverticulum.

Figure S2. Contrast-enhanced computed tomography image showing the esophageal diverticulum near the tracheal bifurcation. (a) Axial image. (b) Coronal image, arrow.

Figure S3. Pathology. (a) Resected specimen. (b) Pathological examination revealing a squamous cell carcinoma confined to the lamina propria mucosae (magnification $\times 100$; H&E stain).

Table S1. Case series of endoscopic submucosal dissection for a superficial esophageal cancer in a diverticulum (in English language only).