Clin J Gastroenterol (2017) 10:442–446
DOI 10.1007/s12328-017-0768-6

CASE REPORT

Distal esophageal spasm with multiple esophageal diverticula successfully treated by peroral endoscopic myotomy

Koji Otani[1] · Shinwa Tanaka[2] · Fumiaki Kawara[2] · Junichi Fujikawa[1] · Akinari Sawada[1] · Risa Uemura[1] · Tetsuya Tanigawa[1] · Toshio Watanabe[1] · Takeshi Azuma[3] · Yasuhiro Fujiwara[1]

Received: 3 March 2017 / Accepted: 25 July 2017 / Published online: 29 July 2017

Abstract Distal esophageal spasm (DES) is a primary esophageal motility disorder. We encountered a rare case of DES accompanied by multiple esophageal diverticula. A 72-year-old woman complained of prolonged dysphagia and chest pain. A barium esophagogram showed multiple esophageal diverticula and significant contraction of the lower esophagus just above the cardia. Esophagogastroduodenoscopy revealed a corkscrew-like appearance, with spiral contractions and diverticula. High-resolution manometry revealed that the integrated relaxation pressure was normal; premature contractions were observed in ≥20% of the swallowing wave; the distal contractile integral was normal. She was diagnosed with DES according to the Chicago classification v 3.0. As smooth muscle relaxants were not effective, we decided to perform peroral endoscopic myotomy (POEM) to eliminate persistent esophageal contraction. After POEM treatment, her symptoms were markedly improved, and the Eckardt score significantly decreased from 11 points to 1. An esophagogram after POEM showed that barium flowed promptly into the stomach. The multiple esophageal diverticula were considered to be the result of false pulsion diverticulosis caused by excessive internal esophageal pressure, and this represented the most severe form of DES. POEM could be a new curative strategy for the most severe DES cases with multiple diverticula.

Keywords Distal esophageal spasm · High-resolution manometry · Chicago classification · Esophageal diverticulum · Peroral endoscopic myotomy

Introduction

Distal esophageal spasm (DES) is a primary esophageal motility disorder, and causes dysphagia and chest pain. Although DES usually presents with a normal peristaltic wave, sustained hypercontraction of the distal esophagus is observed at times. The Chicago classification defines DES as the presence of premature esophageal contractions with normal relaxation of the lower esophageal sphincter, based on high-resolution manometry (HRM). Although esophageal diverticulosis can occur in association with various esophageal motility disorders, there are few reports of DES in association with multiple esophageal diverticula. We report a rare case of symptomatic DES with multiple esophageal diverticula, for which peroral endoscopic myotomy (POEM) was effective.

Case report

A 72-year-old woman had a 5-year history of dysphagia and chest pain, and she experienced episodes of immediate regurgitation of swallowed food. Physical examination was unremarkable. A barium esophagogram showed multiple esophageal diverticula, significant contraction of the lower esophagus just above the cardia, with dilatation of the oral

Koji Otani and Shinwa Tanaka contributed equally to this work.

✉ Koji Otani
kojiotani@med.osaka-cu.ac.jp

[1] Department of Gastroenterology, Osaka City University Graduate School of Medicine, 1-4-3 Asahimachi, Abeno-ku, Osaka 545-8585, Japan

[2] Department of Endoscopy, Kobe University Hospital, Kobe, Japan

[3] Department of Gastroenterology, Kobe University Graduate School of Medicine, Kobe, Japan

esophagus, and pooling of barium at the lower esophagus (Fig. 1a). Esophagogastroduodenoscopy (EGD) revealed multiple esophageal diverticula (Fig. 2a) and a corkscrew-like appearance with spiral contractions from the mid- to lower esophagus (Fig. 2b). HRM was performed to classify the esophageal motility disorder, using the Starlet solid-state system (Star Medical, Inc., Tokyo, Japan), which has 36 circumferential sensors spaced at 1-cm intervals. HRM revealed that the integrated relaxation pressure (IRP) was normal; premature contractions, defined as distal latency (DL) <4.5 s from upper esophageal sphincter relaxation to the contractile deceleration point (CDP), were observed in ≥20% of the swallowing wave; the distal contractile integral (DCI) was within normal limits (1773 mmHg/s/cm) (Fig. 3a).

The patient was diagnosed with DES according to the Chicago classification v 3.0. She was treated with nitroglycerine aerosol, but her dysphagia worsened and it became difficult for her to eat. We then prescribed nifedipine (20 mg/day), isosorbide dinitrate (20 mg/day), and shakuyaku-kanzo-to (traditional Chinese medicine, 7.5 mg/day); her symptoms improved at first, and she was able to eat. However, she again developed dysphagia. We therefore decided to perform POEM, a new minimally invasive endoscopic treatment intended to provide a permanent cure for achalasia and other spastic esophageal motility disorders.

POEM was performed in the operating room under general anesthesia. EGD was performed using a standard scope with CO_2 insufflation. After local injection of normal saline under the esophageal mucosa (Fig. 4a), the surface of the lifted esophageal mucosa was incised (Fig. 4b), and an EGD scope with a plastic cap was inserted under the mucosa. A tunnel was created using endoscopic submucosal dissection with the 3-mm-long type of FlushKnife BT (DK2618JB, Fujifilm, Japan) proximal to the upper border of endoscopically confirmed abnormal esophageal contraction, and was extended 3 cm onto the gastric cardia across the esophagogastric junction (Fig. 4c). A diverticulum without a muscular layer was partially confirmed during the creation of the submucosal tunnel (Fig. 4d). Proximal-to-distal myotomy was subsequently performed from the upper esophagus at 22 cm from the incisors to the gastric cardia at 43 cm from the incisors, with preservation of the longitudinal muscles (Fig. 4e). Finally, the mucosal incision was closed by clipping (Fig. 4f). There were no complications during the intraoperative and postoperative periods, and she was discharged on the 4th postoperative day.

An esophagogram after POEM showed that barium flowed promptly into the stomach (Fig. 1b). Her symptoms were markedly improved, and the Eckardt symptom score, which assesses weight loss, dysphagia, chest pain, and regurgitation, improved from 11 points to 1. HRM performed after POEM showed that esophageal peristalsis had disappeared due to incision of the muscular layer (Fig. 3b). She has had no dysphagia or vomiting since then.

Discussion

Although some esophageal diverticula are associated with esophageal motility disorders, DES in association with multiple diverticula is rare, and is suspected to be caused

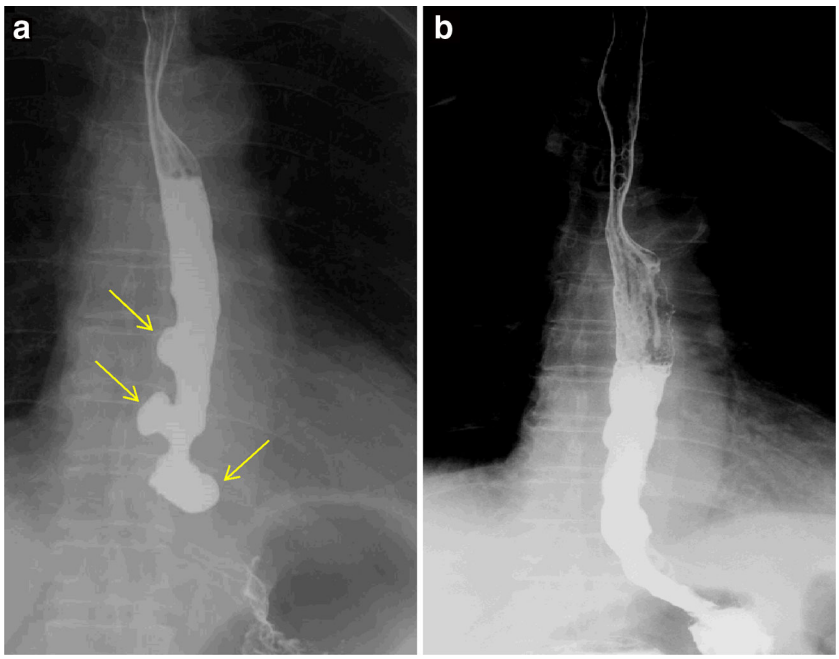

Fig. 1 Esophagograms before and after peroral endoscopic myotomy (POEM). **a** Multiple esophageal diverticula caused by distal esophageal spasm (*arrows*) and significant contraction of the lower esophagus above the cardia before POEM. **b** Smooth inflow of barium into the stomach after POEM

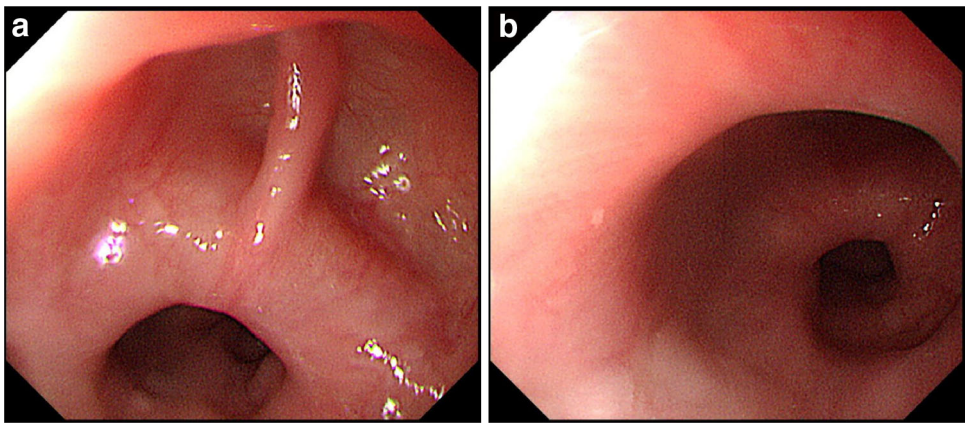

Fig. 2 Endoscopic images of an esophageal diverticulum and corkscrew-like appearance from the mid- to lower esophagus. **a** Esophageal diverticulum caused by distal esophageal spasm. **b** Corkscrew-like appearance with spiral contractions

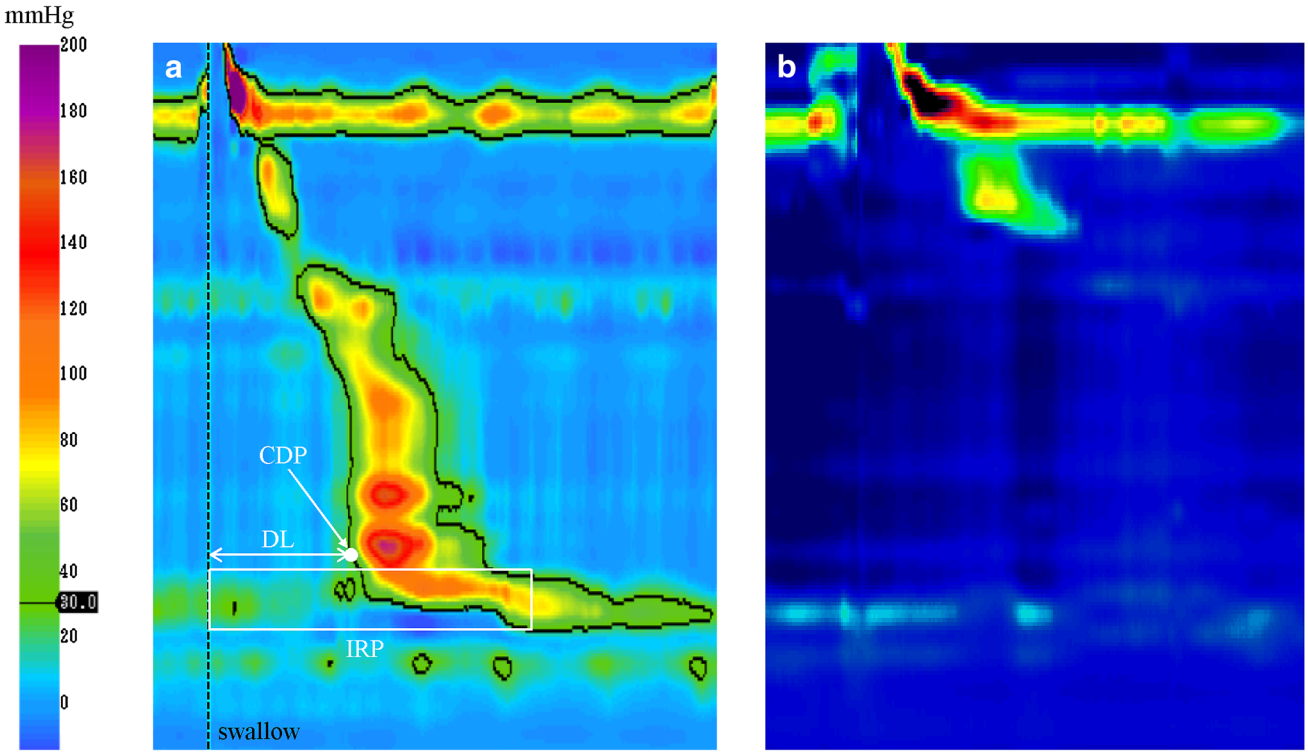

Fig. 3 High-resolution manometry before and after peroral endoscopic myotomy (POEM). **a** Integrated relaxation pressure (IRP) and distal latency (DL) from upper esophageal sphincter relaxation to contractile deceleration point (CDP) of distal esophageal spasm before POEM. **b** Absent esophageal peristalsis after POEM

by excessive esophageal pressure. For the definitive diagnosis of DES, the measurement of esophageal pressure is essential, and the Chicago classification criteria define esophageal motility disorders based on parameters measured by HRM. According to the Chicago classification v 3.0, DES is included in the major disorders of peristalsis and can be diagnosed in cases with premature contraction in ≥20% of the swallowing wave, in combination with a normal median IRP [1]. Premature contraction is defined as DL <4.5 s, and the DL is measured as the interval from the onset of the upper esophageal sphincter relaxation to the CDP, which is the distal esophageal point characterized by an abrupt slowing of deglutitive contraction [2].

The cause of DES is unclear, and gastroesophageal reflux disease (GERD) or stress is suspected to cause the spasm. Proton pump inhibitors are effective for some DES cases caused by GERD [3]. Nitric oxide, calcium channel blockers, phosphodiesterase-5 inhibitors [4], and shakuyaku-kanzo-to have smooth muscle relaxant effects. Tricyclic antidepressants, selective serotonin reuptake inhibitors, and serotonin–norepinephrine reuptake inhibitors are used for the symptom of chest pain. Botulinum

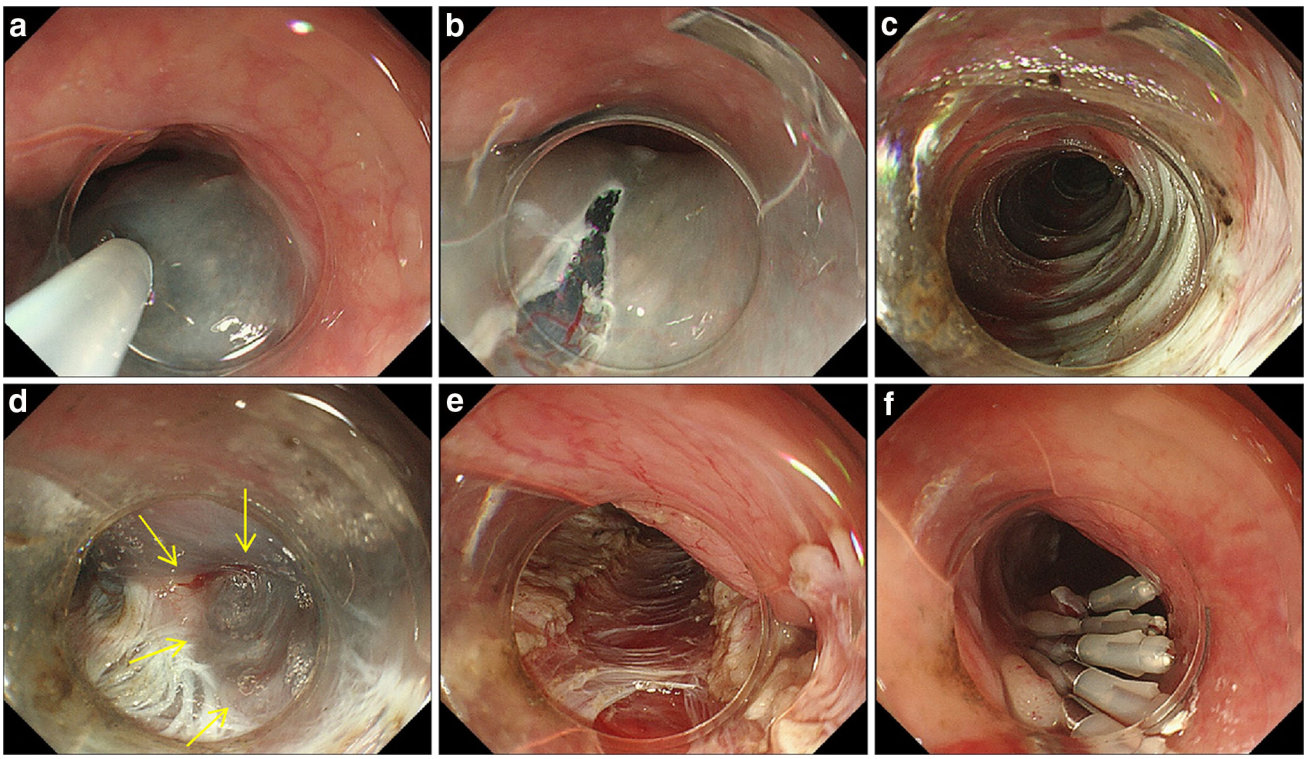

Fig. 4 Procedure of peroral endoscopic myotomy. **a** Local injection of normal saline under the esophageal mucosa. **b** Mucosal incision. **c** Creation of a submucosal tunnel using endoscopic submucosal dissection. **d** A diverticulum without a muscular layer (*arrows*). **e** Proximal-to-distal myotomy, with preservation of longitudinal muscle. **f** Closure of the mucosal incision by clipping

toxin injection of the esophageal body is reported to be useful for relieving symptoms associated with DES [5]. However, the effects of these treatments are palliative, and curative treatment is required. Surgical myotomy for severe esophageal contractions is commonly performed for DES. As minimally invasive and curative treatment to eliminate persistent esophageal contraction is preferred, POEM may be considered as a promising alternative treatment. Inoue et al. developed and established the novel technique of POEM, and positive outcomes were reported in patients with achalasia [6]. POEM has been accepted by endoscopists and surgeons since this report. In addition to achalasia, the indication for POEM has been extended to treat the other types of esophageal motility disorders [7].

We confirmed the loss of the muscle layer in the diverticular portion of the lower esophagus during the POEM procedure. The wall of the lower esophagus is considered congenitally fragile, and a false pulsion diverticulum can be formed by excessive internal esophageal pressure. Extended elevation of esophageal pressure due to the esophageal motility disorder leads to multiple diverticula. DES accompanied by esophageal diverticula is considered the most severe form. Only four case reports of DES with esophageal diverticula were found in PubMed. Matsumoto et al. reported a case with an epiphrenic diverticulum associated with DES [8]. Melman et al. reported that 2 of 11 patients with esophageal motor disorders and epiphrenic diverticula had DES and were treated with a laparoscopic esophageal diverticulectomy and an anterior esophageal myotomy [9]. Vincentine et al. evaluated nine patients with epiphrenic diverticula and found that one DES case was among these [10]. Diverticulectomy and long myotomy has been recommended for patients with such cases to date, as a false pulsion diverticulum is induced by high intraluminal esophageal pressure. This is the first report of DES with esophageal diverticula successfully treated with POEM.

POEM could be a new curative strategy for the most severe DES cases with multiple diverticula, and long-term follow-up to evaluate curative effect will be necessary in the future.

Compliance with ethical standards

Conflict of interest: The authors declare no conflicts of interest for this article.

Human/animal Rights: All procedures followed have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Informed Consent: Informed consent was obtained from the patient in the study.

References

1. Kahrilas PJ, Bredenoord AJ, Fox M, Gyawali CP, Roman S, Smout AJ, et al. The Chicago Classification of esophageal motility disorders, v3.0. Neurogastroenterol Motil. 2015;27(2):160–74.
2. Pandolfino JE, Roman S, Carlson D, Luger D, Bidari K, Boris L, et al. Distal esophageal spasm in high-resolution esophageal pressure topography: defining clinical phenotypes. Gastroenterology. 2011;141(2):469–75.
3. Crozier RE, Glick ME, Gibb SP, Ellis FH Jr, Veerman JM. Acid-provoked esophageal spasm as a cause of noncardiac chest pain. Am J Gastroenterol. 1991;86(11):1576–80.
4. Fox M, Sweis R, Wong T, Anggiansah A. Sildenafil relieves symptoms and normalizes motility in patients with oesophageal spasm: a report of two cases. Neurogastroenterol Motil. 2007;19(10):798–803.
5. Vanuytsel T, Bisschops R, Farre R, Pauwels A, Holvoet L, Arts J, et al. Botulinum toxin reduces dysphagia in patients with nonachalasia primary esophageal motility disorders. Clin Gastroenterol Hepatol. 2013;11(9):1115–1121.e2.
6. Inoue H, Minami H, Kobayashi Y, Sato Y, Kaga M, Suzuki M, et al. Peroral endoscopic myotomy (POEM) for esophageal achalasia. Endoscopy. 2010;42(4):265–71.
7. Hoppo T, Thakkar SJ, Schumacher LY, Komatsu Y, Choe S, Shetty A, et al. A utility of peroral endoscopic myotomy (POEM) across the spectrum of esophageal motility disorders. Surg Endosc. 2016;30(1):233–44.
8. Matsumoto H, Kubota H, Higashida M, Manabe N, Haruma K, Hirai T. Esophageal epiphrenic diverticulum associated with diffuse esophageal spasm. Int J Surg Case Rep. 2015;13:79–83.
9. Melman L, Quinlan J, Robertson B, Brunt LM, Halpin VJ, Eagon JC, et al. Esophageal manometric characteristics and outcomes for laparoscopic esophageal diverticulectomy, myotomy, and partial fundoplication for epiphrenic diverticula. Surg Endosc. 2009;23(6):1337–41.
10. Vicentine FP, Herbella FA, Silva LC, Patti MG. High resolution manometry findings in patients with esophageal epiphrenic diverticula. Am Surg. 2011;77(12):1661–4.