Esophageal epiphrenic diverticulum associated with diffuse esophageal spasm

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ABSTRACT

INTRODUCTION: Esophageal diverticulum, a relatively rare condition, has been considered to be associated with motor abnormalities such as conditions that cause a lack of coordination between the distal esophagus and lower esophageal sphincter.

PRESENTATION OF CASE: We herein report a case of esophageal epiphrenic diverticulum associated with diffuse esophageal spasm. A 73-year-old woman presented with dysphagia and regurgitation. Imaging examinations revealed a right-sided esophageal diverticulum located about 10 cm above the esophagogastric junction. High-resolution manometry revealed normal esophageal motility. However, 24-h pH monitoring revealed continuous acidity due to pooling of residue in the diverticulum. An esophageal epiphrenic diverticulum was diagnosed and resected thoracoscopically. Her dysphagia recurred 2 years later. High-resolution manometry revealed diffuse esophageal spasm.

DISCUSSION: The diverticulum in the present case was considered to have been associated with diffuse esophageal spasm. The motility disorder was likely not identified at the first evaluation.

CONCLUSION: In this case, the patient's symptoms spontaneously resolved without any treatment; however, longer-term follow-up is needed.

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1. Introduction

Epiphrenic diverticulum is a relatively rare condition. It is thought to be a type of pulsion diverticulum and is usually located in the distal 10 cm of the esophagus [1]. Symptoms include dysphagia, odynophagia, regurgitation, chest pain, heartburn, and/or aspiration. The size of the diverticulum may influence the severity of symptoms [2]. More recently, esophageal diverticula have been considered to be associated with motor abnormalities [1–8], such as conditions that cause a lack of coordination between the distal esophagus and lower esophageal sphincter (LES) [3].

We experienced a patient with an esophageal epiphrenic diverticulum with no apparent preoperative motor abnormality. However, high-resolution manometry (HRM) revealed diffuse esophageal spasm (DES) when she developed symptom recurrence 2 years postoperatively.

2. Presentation of case

A 73-year-old woman was referred to our hospital in November 2009 for evaluation of a 7-month history of dysphagia and food regurgitation. She underwent endoscopy, upper gastrography, and computed tomography (Fig. 1), which revealed a diverticulum located on the right side of the esophagus about 10 cm above the esophagogastric junction. HRM revealed an LES pressure of 7 mmHg, residual pressure of 0 mmHg upon LES relaxation, and normal contraction (InSIGHT G3; Sandhill Scientific, Inc., Denver, CO) (Fig. 2a). However, according to her 24-h pH monitoring results, the fraction of time that the pH was <4 was higher than normal at 24.5% (Digitrapper pH 400; Medtronic, Minneapolis, MN) (Fig. 3a).
She was diagnosed with a mid-esophageal epiphrenic diverticulum. On 2 November 2009, thoracoscopic diverticulectomy via the right side was performed using a stapler (Endo GIA Tri-Staple™; Covidien, Mansfield, MA), and the staple line was covered with esophageal muscle using intermittent 4–0 Biosyn™ (Covidien) sutures. There was no adhesion between the diverticulum and lymph nodes around the tracheobronchus. Her postoperative course was uneventful. Her regurgitation improved, and the fraction of time that the pH was <4 was 0% according to her 24-h pH monitoring results (Fig. 3b). She was discharged on 21 November 2009.

On 6 June 2013, she presented again to our hospital because of recurrent dysphagia. She underwent HRM re-examination of her esophageal motility (Fig. 2b). The distal contractile integral was high at 6533 mmHg/s/cm; however, it did not reach 8000 mmHg/s/cm, a diagnostic criterion of hypercontractile esophagus. More than 20% premature contractions was observed, indicating DES characterized by hypertensive peristalsis of the esophagus. Endoscopic ultrasonography was performed to evaluate the esophageal wall. The wall was thick at >1 cm, which was caused by the esophageal hypercontractility. The diverticulum was suspected to have been caused by the high pressure induced by the DES. Long myotomy and fundoplication were recommended; however, her dysphagia improved 1 month after the HRM examination. At the time of this writing, she was continuing to undergo follow-up.

### 3. Discussion

An epiphrenic diverticulum is a false pulsion diverticulum that is considered to be caused by esophageal motor abnormalities such as achalasia, DES, hypertensive LES, nutcracker esophagus, or nonspecific esophageal motility disorders. Epiphrenic diverticula are most frequently associated with achalasia [1–8]. When the achalasia is the cause of the diverticulum, surgical treatment comprising diverticulectomy, myotomy, and fundoplication is required [1–7]. This treatment is generally performed by a laparoscopic approach [4,8].

Another cause of epiphrenic diverticula is DES [2,4,5]. Melman et al. [2] reported the presence of esophageal body motor disorders and/or LES abnormalities in 11 patients with epiphrenic diverticula: 3 patients had achalasia, 1 had vigorous achalasia, 2 had diffuse esophageal spasm, and 5 had a nonspecific motor disorder. Vincente et al. [4] reported using HRM to determine the cause of nine cases of epiphrenic diverticulum and found achalasia in six, nonspecific disorders in two, and DES in one. In their study, HRM demonstrated motor abnormalities in patients with esophageal epiphrenic diverticulum.

In the present case, relaxation of the LES was normal before the operation, and no other abnormalities were detected. For this reason, we performed diverticulectomy via thoracoscopy only, and a good result was obtained. Two years postoperatively, DES was diagnosed based on the presence of esophageal hypercontractility.
Fig. 2. Twenty-four-hour pH monitoring results. (a) Preoperative 24-h pH monitoring showed continuous acidity secondary to the pooling of residue within the diverticulum. (b) Postoperative 24-h pH monitoring results were normal.

on HRM. Some authors have recommended diverticulectomy and myotomy for epiphrenic diverticula even if not caused by achalasia [1,2,4–8] because an epiphrenic diverticulum is thought to be a type of pulsion diverticulum induced by high intraluminal pressure in the lower esophagus. Others have advocated the selective use of myotomy only in patients with evident hypertonic esophageal motility based on motility test results [3].

Based on HRM findings, the diverticulum in the present case was considered to have been associated with DES. The motility disorder was likely not identified at the first evaluation. Not all patients with DES exhibit the same clinical course and some cases spontaneously resolve [9]. In this case, the patient’s symptoms resolved without any treatment; however, longer-term follow-up is needed.

4. Conclusion

Clinicians must not forget the possibility of esophageal motility disorder in the treatment of diaphragmatic diverticulum.

Conflict of interest

None.
Fig. 3. High-resolution manometry findings. (a) Normal motility was revealed before the diverticulectomy. (b) Peristaltic waves were present in the esophageal body. Fully synchronized waves of contraction occurred with marked increases in the esophageal body pressure (257 mmHg) and lower esophageal sphincter residual pressure (5.9 mmHg; normal, <8 mmHg). The distal contractile integral was very high at 6533 mmHg/s/cm. More than 20% premature contractions was observed.
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Ethical approval

This is not the research study, so that the ethical approval was not given.

Consent

Informed consent has been obtained.

Author's contribution

H. Matsumoto was correspondent author, H. Kubota and M. Higashida were the main treating physician, N. Manabe and K. Haruma were contribute to diagnosis, and T. Hirai was the representative of our department.

Guarantor

Hideo Matsumoto.

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