Recurrent Esophageal Stricture as a Result of Esophageal Diverticulum: Case Report

Kaka Renaldi, Muhamad Reza Prabowo***

*Division of Gastroenterology, Department of Internal Medicine
Faculty of Medicine, Universitas Indonesia/Dr. Cipto Mangunkusumo General National Hospital, Jakarta
**Department of Internal Medicine, Faculty of Medicine
Universitas Indonesia/Dr. Cipto Mangunkusumo General National Hospital, Jakarta

Corresponding author:
Muhamad Reza Prabowo. Department of Internal Medicine, Dr. Cipto Mangunkusumo General National Hospital. Jl. Diponegoro No. 71 Jakarta Indonesia. Phone: 021-1500135. E-mail: muhammadrezaprabowo@gmail.com

ABSTRACT

Esophageal stricture is a disorder that limiting patients' capability to get adequate intake. Dysphagia and regurgitation are main problem that make patient admit to the hospital. There are several causes that narrowing esophageal lumen; those could be intraluminal or extraluminal. Esophageal diverticulum is one of a kind. It is not easy to establish the diagnosis of diverticulum in esophagus segment since the supporting examinations needed were complex. Esophageal diverticulum changes the anatomy of esophagus itself so the musculature function would be interfered. Esophageal stricture treatment should be interdisciplinary approach because inappropriate management would increase risk of complication and lowering patient's quality of life. The management of esophageal diverticulum can be challenging and its prognosis depends on the patient's characteristic and comorbidity.

Keywords: *dysphagia, esophageal stricture, esophageal diverticulum*

ABSTRAK

Striktur esofagus merupakan suatu kondisi kelainan saluran cerna yang dapat menyebabkan penurunan kemampuan asupan pasien yang adekuat. Disfagia dan regurgitasi merupakan alasan pasien berobat ke rumah sakit. Terdapat beberapa penyebab menyempitnya lumen esofagus, baik intralumen ataupun ekstralumen. Divertikulum esofagus merupakan salah satunya. Tidak mudah mendiagnosis divertikulum pada esofagus karena membutuhkan pemeriksaan penunjang yang mumpuni. Divertikulum esofagus menyebabkan perubahan struktur anatomi esofagus itu sendiri sehingga menyebabkan perubahan fungsi otot yang ada pada esofagus. Penatalaksanaan striktur esofagus pada divertikulum esofagus memerlukan penanganan secara interdisiplin karena penatalaksanaan yang tidak sesuai dapat menyebabkan komplikasi dan penurunan kualitas hidup pada pasien. Tata laksana divertikulum esofagus tidaklah mudah dan prognosisnya bergantung karakteristik dan komorbiditas pasien itu sendiri.

Kata kunci: *disfagia, striktur esofagus, divertikulum esofagus*

INTRODUCTION

Esophageal stricture is an abnormal condition that narrowing the esophageal lumen. Patients almost always come to the doctor with complaints of difficulty swallowing. There are so many etiologies which cause esophageal stricture.[1] Benign conditions include corrosive material ingestion, pill-induced esophagitis, diverticulum, eosinophilic esophagitis, post-surgery, injury (radiation, chemotherapy, thermal), infection (cytomegalovirus/CMV, herpes simplex, human immunodeficiency virus/HIV, candidiasis), and iatrogenic.[2–5] Meanwhile esophageal adenocarcinoma, squamous cell carcinoma, and metastatic neoplasms (usually from lung cancer) are malignant causes of this situation.[1]

Clinicians also missed other rare causes such as prolonged use of nasogastric tube, collagen disease, Crohn's disease, and tuberculosis.[6,7] The incidence increases accordance with age and the most widely risk factors are history of gastroesophageal reflux disease (GERD), peptic ulcer disease, and alcohol consumption.[8]

Evaluation of esophageal stricture needs history taking, physical examination, and also effective supporting examination. Upper gastrointestinal (UGI) endoscopy is the most important intervention in the case of stricture.[9] Diverticulum sometimes may not be detected, additional imaging may help the diagnosis.[10] Any stricture (including diverticula) requires therapy to make sure luminal patency. Some techniques are performed to achieve that goal, like medical management, dilators, stent, and resection.[11] The main goal is relieving dysphagia which may lead to improved quality of life.[12]

CASE ILLUSTRATION

A 62-year-old man came to fast-track ward of Dr. Cipto Mangunkusumo General National Hospital with a chief complaint of difficulty in swallowing 3 months prior to admission. He had been planned to have dilatation procedure per endoscopy since his last visit at outpatient clinic. At first, he had nausea and vomiting once a week on 2017. There was no epigastric pain, regurgitation, and heartburn. Dysphagia started in 2019, firstly he could not swallow solid food and then he was difficult to swallow semisolid material. That was the first time he visited the doctor.

Since then, he often vomited so the appetite getting decreased, and he also lost 10 kg in 2 years. He came to gastroenterology clinic and was planned to perform esophagogastroduodenoscopy (EGD) and endoscopic ultrasound-fine needle aspiration (EUS-FNA). Because there were superficial and deep esophageal varices, the operator did not perform the FNA. Because semisolid food could not be ingested, the nasogastric tube was placed twice in 3 months as an enteral access.

The interdisciplinary meeting was held after thorax computerized tomography (CT) result concluded. At the end of discussion, the surgical option was chosen since esophageal diverticulum was suspected. Around six months after meeting (January 2020), esophagectomy was performed by digestive and thoracic surgeon. Sample of the esophagus was taken and the results showed inflammatory esophagus in accordance with the esophageal diverticulum. The patient was hospitalized for one month and was diagnosed as relapse pulmonary tuberculosis (TB) because lung bullae and fibroinfiltrate were accidentally found in thorax CT and chest X-ray. The patient had decreased appetite and weight loss. There were no any chronic cough, fever, dyspnea, or night sweat. Streptomycin injection was given additional to standard TB treatment.

Six months after surgery (July 2020), he came back with same problem. He only could swallow liquid food and his body weight diminished approximately 5 kg. The clinicians decided to perform dilatation per UGI endoscopy. Successful procedure was attempted twice in 3 months (October–December 2020), but he never came back in two years because of Coronavirus disease 2019 (COVID-19) pandemic. At the same time, he finished TB medication for 12 months and was declared cured from the CT evaluation.

He had difficulty swallowing in 3 months prior admission (May 2022), so he came back to the gastroenterology clinic. He lost 5 kg in 2 years. He denied any epigastric pain, heartburn, regurgitation, hematemesis, melena, or growling abdomen. There was no lump palpable. He also denied history of drug consumption, corrosive material ingestion, or suicidal attempt. There was no hypertension and diabetes mellitus. His daily meals are blended food, fruit juice, and milk. He did not smoke, consume alcohol, use IV drug and tattoo, and also promiscuity. There was no radiation and chemotherapy exposure. He was a retired entrepreneur and using government insurance.

On the physical examination, he looked mildly ill and fully awake. His body weight and height were 53 kg and 163 cm. His body mass index was 19.94 kg/m^2. His vital signs were stable. He had postoperative scar on his chest and upper abdomen. No other abnormal condition was observed.

From the laboratory examination, it was found that hemoglobin was 14.3 g/dL, hematocrit 40.7%, leukocyte 6,960 cells/μL, and platelet 305,000 cells/μL. His prothrombin time (PT) and activated partial thromboplastin time (aPTT) were 10 (control 11.4) and 27.6 (control 34.2). Sodium, potassium, and chloride level were 139, 4.2, and 103.9 mEq/L respectively. Albumin was 4.5 g/dL. Ureum and creatinine were 23.5 mg/dL and 1 mg/dL. Estimated glomerular filtration rate (GFR) was 80.3 mL/min/1.73 m^2. Random blood glucose was 53 mg/dL.

After performing dilatation technique, the routine blood test was conducted. It was found that hemoglobin was 14.3 g/dL, hematocrit 42%, leukocyte 8,160 cells/μL, and platelet 281,000 cells/μL.

Figure 1. Multiple bullae on thorax CT

Several radiology examinations were used to help the diagnosis. First thorax CT (with contrast, as shown in Figure 1 showed heterogeneous circular thickening of esophageal wall from esophagogastric junction (EGJ) from Th2-Th12, with irregular border, lobulated and diffuse margin, and dilated proximal esophagus. There were fibrosis, bronchiectasis, and infiltrate in bilateral segment of the lungs, and also multiple bullae in both of lungs suggesting pulmonary TB. Chest CT was repeated 2 years later and indicated hiatal hernia and dilated proximal esophagus, settled multiple bullae in both of lungs, and fibrosis, bronchiectasis, tuberculoma of the apex. The chest x-ray also supported that there were fibroinfiltrate in both of lungs.

Upper abdominal CT with contrast showed pulsion diverticulum at the distal esophagus (epiphrenic diverticulum) and thickening of distal esophageal wall to EGJ. There was visualized nodule on lung bases.

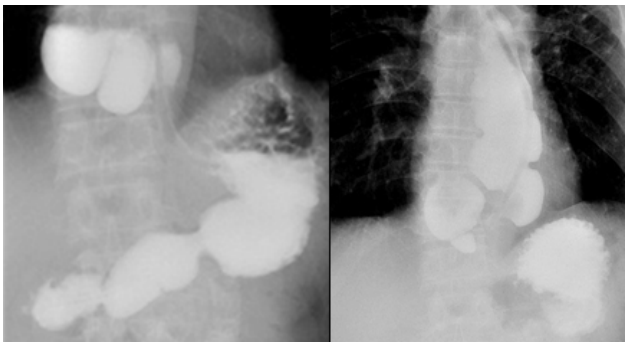

Figure 2. Filled contrast in outpouching right to distal esophagus suggesting diverticulum

Barium swallow was attempted before surgery, defining luminal dilatation, and corkscrew esophagus suggesting diffused esophageal spasm as differential diagnosis. There was outpouching filled with contrast right to the distal esophagus suggesting esophageal diverticulum, and EGJ luminal narrowing (Figure 2).

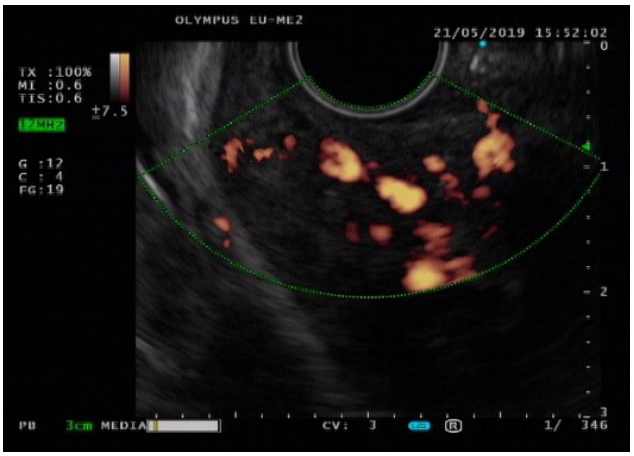

Figure 3. Multiple varices in esophageal EUS

Endoscopy procedure was done multiple times. At first, endoscopic ultrasound (EUS) showed superficial and deep varices form distal esophagus to EGJ (Figure 3), hence the fine needle aspiration (FNA) was not performed. Meanwhile UGI endoscopy showed dilatation and sigmoidization of esophagus, two giants epiphrenic diverticulum were showed on EGJ, pangastritis, and multiple antrum ulcer (Figure 4). Next EGD was performed to dilate the esophagus by using balloon technique, with 12–13.5 mm in diameter and 6–8 atm in pressure. Dilatation was attempted 20 cm from incisivus (distal esophagus).

Histopathology preparate from EGD always showed chronic gastritis without any dysplasia or atrophy. From intraoperative biopsy taken two years ago, the sample figured reflux esophagitis with stricture and diverticulum besides active chronic gastritis.

There was no malignancy or infection (granuloma, caseous necrosis, etc) stated in microscopic findings. The meeting concluded that the stricture came from the esophageal diverticulum.

From the anamnesis, physical examination, and supporting tests, the patient was assessed as esophageal stricture, gastritis with gastric ulcer, malnutrition, and complete medication of relapse TB. The patient was informed to have dilatation per UGI endoscopy every 2–4 week, double dose proton pump inhibitor (PPI), and oral nutrition (semisolid food, soft texture, no irritated material). Till nowadays, the EGD with dilatation has been performed five times.

The patient came home safely with uneventful post-procedural complication. He can ingest semisolid food and come to visit gastroenterology clinic to observe his symptoms routinely. He feel much better after successful dilatation procedure.

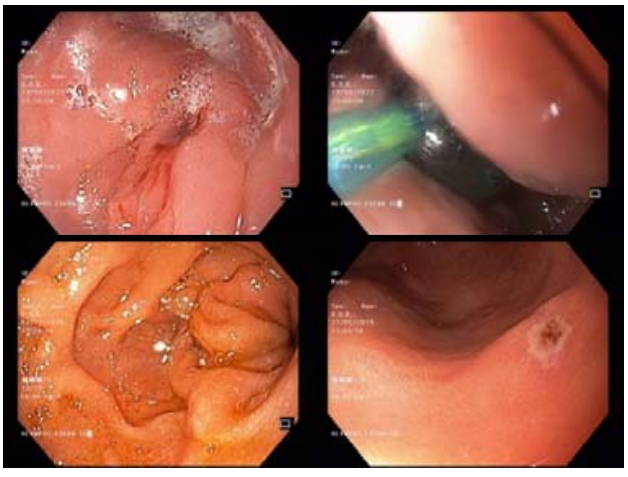

Figure 4. (A) Esophageal stricture; (B) Balloon dilatation; (C) Gastritis; (D) Antrum ulcer

DISCUSSION

Esophageal stricture can occur in any population, regardless of age. Caustic injury and eosinophilic esophagitis are more common in younger patients, while acid reflux, pill-induced, and iatrogenic are more frequent in adult population. Tuberculosis as infection agent which could be manifested in many organs including esophagus, should be considered as differential diagnosis of esophageal stricture.[8] Moreover, malignancy and diverticula are more prevalent in older population. They are generally found more in men.[13]

The patient is a 62-year-old male, his age at esophageal stricture diagnosis was 59 years old. From this age group, we can simplify the etiology of stricture would be either diverticulum or malignancy prior to additional examination that could establish the working diagnosis. Clinical manifestations of stricture esophagus include dysphagia, odynophagia, weight loss, and meal impaction. Progressive dysphagia is hallmark of esophageal stenosis.[14]

This patient had difficulty in swallowing solid food at first, and then semisolid food. He had lost total 25 kg in 5 years. These symptoms showed compatibility to esophageal stricture regardless of its etiology since inadequacy of intake would make patient getting malnourished.

Upper GI endoscopy is essential in diagnostic and management of a stricture. Moreover, biopsy to the stricture may confirm its etiology and peptic stricture could be further investigated.[9] The biopsy of this case showed the diverticulum of esophageal tissue therefore biopsy has vital role for this entity. Barium swallow will help to define the level, size, length, and severity of stricture, particularly in total occlusion.[10] EUS provide more information since its high-resolution images, hence it has benefit to differentiate malignant and benign stricture from its wall. Thorax CT may also determine the size and extent of extraluminal abnormality.[11] Esophageal dilatation resulted from Giant epiphrenic diverticulum on esophagus were visible in UGI endoscopy. Pangastritis and antrum ulcer are evidence that peptic stricture was involved to aggravate beside the esophageal diverticulum as shown in histopathology and imaging examination. Barium swallow also showed luminal dilatation and contrast-filled outpouching at distal esophagus region. Additionally, both thorax and abdominal CT scan defined the esophageal wall thickening suggesting epiphrenic diverticulum. Unfortunately, FNA was not performed since multiple varices shown in the EUS.

Some techniques were used to achieve luminal patency. Mechanical dilatation using balloon expansion produced radial force. Recurrence is defined as esophageal diameter less than 14 mm after dilatation was done in 4 weeks.[15] Adjunctive steroid injection could decrease inflammation and reduce restenosis incidence.[16] Esophageal stents might be chosen in case of malignant stricture for complete palliation.[17] Last but not least, surgical resection is necessary for malignant disorder or benign structure that refractory to medical/endoscopic therapy. Enteral tube placement like nasogastric tube (NGT) could provide enteral nutrition if the stricture was incomplete.[18]

Initially, the NGT was inserted twice to make access for nutrition prior definitive treatment given. However,

after tight discussion within the team, the patient underwent esophagectomy before any endoscopic therapy was attempted. This was due to clinical findings that point to malignancy (quick progressive dysphagia and weight loss). Despite surgery had been performed, the patient had recurrent dysphagia so he must have had gradual dilatation per endoscopy with balloon technique. One reason that would make it possible is post-surgery inflammation. It seemed first dilatation was successful because there was improvement in diameter length on next endoscopy (12–13.5 mm), yet the patient did not come to have third dilatation since the pandemic situation. He came back with the same size (12 mm) diameter. Steroid injection was not given because of relapse pulmonary TB established on thorax CT (multiple bullae). From this statement above, dilatation could be the first line treatment and surgery is alternative option to recurrent esophageal stricture since surgery has also chance of relapse afterward.

Gold standard for the diagnosis of esophageal stricture is histopathology. Benign and malignant condition could be differentiated carefully from any samples. Inflammatory changes as a pathophysiology basis of esophageal stricture could be starting point for the development of fibrosis which led to luminal stenosis.[19] Malignant stricture could be derived from esophageal adenocarcinoma, squamous cell carcinoma, or metastatic neoplasm.[1]

Chronic gastritis from EGD samples defined peptic stricture contribute to esophageal constriction. Intraoperative biopsy supported that situation by showing reflux esophagitis. Another vital statement indicated the diverticulum as etiology of the stricture. Neither infection (like TB as experienced by the patient) nor malignancy was found in the preparate.

Corkscrew appearance in barium swallow might confuse the clinician to diagnose the patient because he had diffuse esophageal spasm but did not experience chest pain. Achalasia could be suspected if only history taking and physical exam were used as diagnostic tools. Nevertheless, barium swallow did not show bird beak appearance. Either diffuse esophageal spasm or achalasia could be eliminated through esophageal manometry.[20,21]

After dilatation or surgical procedure, patient will have 10–30% chance of restenosis. Therefore, some strategies were made to minimize the probability. Long term PPI therapy beside dilatation might decline the restenosis incidence in case of peptic stricture.[22] Steroid injection could decrease stricture recurrence by suppressing the inflammation.[16] Double dose omeprazole was given daily to this patient since gastritis also found in the stomach. Beneath this condition, steroid injection per endoscopy is still not given nowadays since the physician concerned of his relapsing TB despite its minimal systemic effect. He also could have semisolid and liquid food better after balloon dilatation.

From the explanation above, we can see that esophageal stricture with rapidly clinical deterioration is not always caused by malignancy. Refractory dysphagia experienced by this patient was caused by esophageal epiphrenic diverticulum that until now the process to form the diverticula is not fully known.[22]

Esophageal diverticulum is divided into pharyngeal (Zenker), mid-esophageal, and epiphrenic. Epinephric diverticulum (pulsion diverticulum) is formed because the distal wall pushed as a result of mismatch between lower esophageal sphingter (LES) and the lower esophagus. Dysphagia is most common symptom, so that it is quite similar from other strictures.[13] In this case, the patient was performed surgery followed by EGD with dilatation. The management of esophageal diverticulum somewhat similar with management of strictures in general. Nowadays, small myotomy and diverticulectomy in addition to endoscopic procedure would be treatment choice for symptomatic patient in the future.[13]

The prognosis itself depends on patient's age and comorbidity. Immediate surgery does not guarantee either improving condition or recurrence rate, even surgical complication can be a poor prognosis for the patient.[23]

From this case we learn that not all stricture with poor clinical condition is caused by malignancy, but benign stricture can also deteriorate clinical symptoms if does not manage properly. Patients' characteristic and comorbidity influence the outcome of the treatment given to the patient.

As a conclusion, esophageal stricture can be caused by many etiologies, one of which is diverticulum. Other participating conditions in the patient can affect the choice of therapy, so careful examination must be addressed to prevent complication. Interdisciplinary approach is needed to give the best service to the patient, not only for patients' health (because of stricture), but also their quality of life.

REFERENCES

1. Desai JP, Moustarah F. Esophageal stricture. StatPearls [serial online] [cited 2022 Aug 25]. Available from: https://www.ncbi.nlm.nih.gov/books/NBK532858/.
2. Mowry JB, Spyker DA, Cantilena LR, McMillan N, Ford M. 2013 Annual report of the American association of poison control centers' national poison data system (NPDS): 31st annual report. Clin Toxicol (Phila) 2014;52:1032–283.
3. Schoepfer AM, Safroneeva E, Bussmann C, Kuchen T, Portmann S, Simon HU, et al. Delay in diagnosis of eosinophilic esophagitis increases risk for stricture formation in a time-dependent manner. Gastroenterology 2013;145:1230–6.
4. Kim SH, Jeong JB, Kim JW, Koh SJ, Kim BG, Lee KL, et al. Clinical and endoscopic characteristics of drug-induced esophagitis. World J Gastroenterol 2014;20:10994–9.
5. Martínek J, Juhas S, Dolezel R, Walterová B, Juhasova J, Klima J, et al. Prevention of esophageal strictures after circumferential endoscopic submucosal dissection. Minerva Chir 2018;73:394–409.
6. Moleiro J, Faias S, Afonso A. Esophageal stricture of an unusual etiology. Clin Gastroenterol Hepatol 2016;14:59–60.
7. Mbiine R, Kabuye R, Lekuya HM, Manyillirah W. Tuberculosis as a primary cause of oesophageal stricture: a case report. J Cardiothorac Surg 2018;13:58.
8. Richter JE, Rubenstein JH. Presentation and epidemiology of gastroesophageal reflux disease. Gastroenterology 2018;154:267–76.
9. Shami VM. Endoscopic management of esophageal strictures. Gastroenterol Hepatol 2014;10:389–91.
10. Luedtke P, Levine MS, Rubesin SE, Weinstein DS, Laufer I. Radiologic diagnosis of benign esophageal strictures: a pattern approach. Radiographics 2003;23:897–909.
11. Sami SS, Haboubi HN, Ang Y, Boger P, Bhandari P, de Caestecker J, et al. UK guidelines on oesophageal dilatation in clinical practice. Gut 2018;67:1000–23.
12. Rana SS, Sharma R, Gupta R. High-frequency miniprobe endoscopic ultrasonography for evaluation of indeterminate esophageal strictures. Ann Gastroenterol 2018;31:680–4.
13. Yam J, Baldwin D, Ahmad SA. Esophageal Diverticula. StatPearls [serial online] [cited 2022 Aug 25]. Available from: https://www.ncbi.nlm.nih.gov/books/NBK532858/.
14. Smith CD. Esophageal strictures and diverticula. Surg Clin North Am 2015;95:669–81.
15. Raymondi R, Pereira-Lima JC, Valves A, Morales GF, Marques D, Lopes CV, et al. Endoscopic dilation of benign esophageal strictures without fluoroscopy: experience of 2750 procedures. Hepatogastroenterology 2008;55:1342–8.
16. Yan X, Nie D, Zhang Y, Chang H, Huang Y. Effectiveness of an orally administered steroid gel at preventing restenosis after endoscopic balloon dilation of benign esophageal stricture. Medicine (Baltimore) 2019;98:e14565.
17. Hindy P, Hong J, Lam-Tsai Y, Gress F. A comprehensive review of esophageal stents. Gastroenterol Hepatol 2012;8:526–34.
18. Pregun I, Hritz I, Tulassay Z, Herszényi L. Peptic esophageal stricture: medical treatment. Dig Dis 2009;27:31–7.
19. Kuo WH, Kalloo AN. Reflux strictures of the esophagus. Gastrointest Endosc Clin N Am 1998;8:273–81.
20. Frieling T. Non-cardiac chest pain. Visc Med 2018;34:92–6.
21. Swanström LL. Achalasia: treatment, current status and future advances. Korean J Intern Med 2019;34:1173–80.
22. Wang ZM, Zhang SC, Teng X. Esophageal diverticulum serves as a unique cause of bronchoesophageal fistula in children: a case report. Medicine (Baltimore) 2017;96:e9492.
23. Yuan Y, Zhao YF, Hu Y, Chen LQ. Surgical treatment of Zenker's diverticulum. Dig Surg 2013;30:207–18.