

Case Report

Surgical versus conservative management for suppurative intramural esophageal dissection

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Abstract

Often presented with an insidious onset, intramural esophageal dissection (IED) is a rare disease which has been mostly treated conservatively, though the treatment for infectious cases is still controversial. Two cases are reported here as follows: one was approached surgically by thoracoscopic mediastinotomy and neck debridement and the other was treated conservatively. The latter subsequently developed septic shock and was readmitted with intensive care. Both patients eventually recovered and resumed oral intake without requiring esophagectomy. Comparing these two cases of IED, one with a mucosal perforation and the other without, surgical intervention revealed the advantages of definitive isolation of pathogen and rapid mechanical removal of pus. Surgical risks should be evaluated against the severity, such as the presence of a mucosal perforation and the risk of a protracted clinical course for IED.

Keywords: Abscess, intramural esophageal dissection, treatment, drainage

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INTRODUCTION

First described in 1968 by Marks and Keet, intramural esophageal dissection (IED) is an uncommon disease of the upper gastrointestinal tract limited to the case reports in the medical literature till date.^[1,2] There are two main types of IED, namely, one that initiates with a mucosal break followed by a submucosal separation along with the length of the esophagus, and the other that stems from underlying coagulopathy with hematoma formation in the submucosal layer of the esophagus followed by a mucosal tear.^[3] We present, herein, two cases of IED, both of which are infectious in nature.

CASE REPORTS

Case 1

A 59-year-old male patient with a history of cerebral infarction, hypertension, diabetes mellitus, and dyslipidemia presented with an intermittent fever with neck pain and swelling for a week, and dysphagia for 1 day. History of foreign-body ingestion, dental problem, or trauma was denied. Laboratory investigation revealed a white blood cell (WBC) count was 20940/ μ L and an elevated C-reactive protein (CRP) of 27.2 mg/dl. Computed tomography (CT) showed an abscess accumulation in the right pyriform sinus and swelling of the entire esophageal wall with intramural air [Figure 1]. Subsequent esophagoscopy

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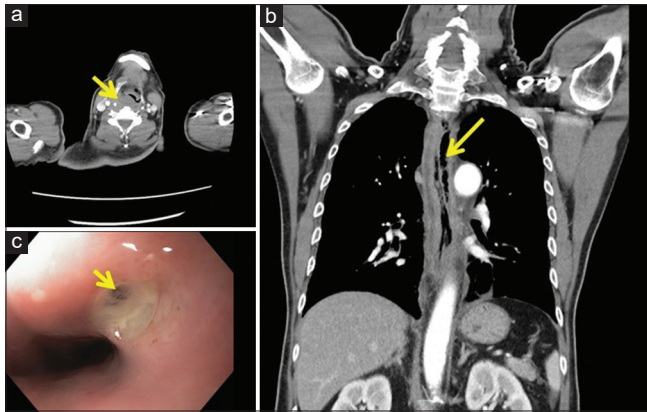


Figure 1: (a) Computed tomography – Abscess in the right pyriform sinus. (b) Computed tomography – Entire esophageal wall was swollen with intramural air. (c) Esophagoscopy showed a right pyriform sinus mucosal perforation with pus

showed a mucosal perforation in the left pyriform sinus draining pus.

Emergent neck debridement, video-assisted thoracoscopic mediastinotomy along with the whole length of thoracic esophagus, and laparoscopic jejunostomy were performed. Intraoperatively, deep neck abscess, purulent pleural effusion, and an edematous esophagus with mural abscess were noted. Much pus was noted and drained from the mediastinal and pleural spaces, but there was no perforation of the thoracic esophagus observed. During the manipulation, about 100 ml pus was also cleared from the perforation of the pyriform sinus mucosa out through the mouth.

After the operation, enteral feeding was implanted, and piperacillin/tazobactam and metronidazole were prescribed. *Klebsiella pneumoniae* was isolated. The patient was discharged 18 days after the surgery. An esophagoscopy 1 month later showed a completely healed mucosa.

Case 2

A 64-year-old male patient with a history of hypertension and diabetes mellitus, presented with a sudden onset of chest pain and epigastralgia after an episodic cough associated with cigarette smoking. In the following days, dysphagia with solid food developed. Foreign-body ingestion was denied. Initial laboratory tests showed a WBC of 15,940/ μ L, an elevated CRP of 26.4 mg/dL. CT showed the presence of submucosa swelling throughout the entire esophagus [Figure 2a]. Esophagogastroduodenoscopy revealed the esophageal mucosal bulging and lumen narrowing without mucosal disruption.

The patient was managed conservatively with nothing by mouth and flomoxef. His clinical symptoms improved and the CRP was trending down. Therefore, he was

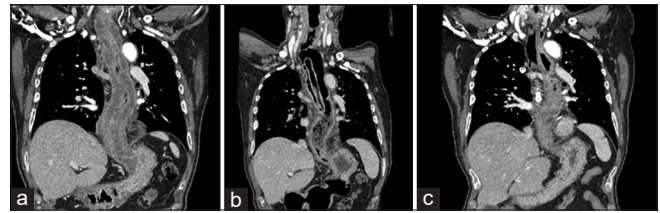


Figure 2: Computed tomography scans. (a) The esophagus with submucosal swelling. (b) The esophagus with submucosal air-fluid and dissection. (c) The reattachment of the mucosa to the muscle layer with minimal residual abscess

discharged 8 days later with amoxicillin/clavulanic acid. On day 15, he presented with recurrent chest pain, fever, and shock. CT revealed submucosal dissection with air surrounding the thin isolated mucosa, and fluid in the lower esophagus [Figure 2b]. He was admitted to the intensive care unit (ICU) under a critical condition with septic shock and was treated with flomoxef. After 25 days of hospital stay including 3 days of ICU stay, CT imaging later showed reattachment of the esophageal mucosa to the muscle layer and decreased air-fluid accumulation within the esophageal submucosa [Figure 2c]. The patient was totally asymptomatic with normal swallowing function during the follow-up.

DISCUSSION

The submucosa of the esophagus is a layer of loose connective tissue with lymphatic network tunneling the entire esophagus that renders it vulnerable to hemorrhage, infection, and then dissection.^[1] Generally afflicting the elderly females, IED is characterized by a mucosa break in the esophagus and a lengthy dissevering space between the mucosa and submucosa of the esophagus.^[2] Etiology of IED ranges from iatrogenic, foreign-body ingestion, infection, bleeding tendency, autoimmune disease, and spontaneous occurrence.^[1]

Clinical presentation characteristic of IED includes chest pain, epigastric pain, radiating back pain, dysphagia, odynophagia, nausea, vomiting, and hematemesis.^[4] Esophagoscopy, esophagography, and CT-scan are the diagnostic tools.

Most of the noninfectious IED have been treated conservatively, but for suppurative IED, antibiotics and drainage were the treatment of choice. Case reports have suggested various drainage approaches including mediastinotomy,^[5] endoscopic intraluminal drainage,^[6-8] and esophagectomy.^[9]

In the first case, the patient had an obvious pyriform sinus mucosa perforation, a deep neck abscess, a phlegmon that

dissected downwardly along with the submucosa of the entire esophagus, penetrated into the mediastinum, and extended into the pleural space. Adequate drainage was administered. In contrast, the second patient who received conservative treatment had a submucosal infection of the whole esophagus which progressed into a phlegmon with severe IED. Septic shock developed despite oral antibiotics use. The patient eventually recovered after a prolonged hospitalization. According to the degree of severity of IED suggested by Skinner and Belsey, esophageal perforation could be classified as follows: limited air leakage only, perforation with self-drainage, free mediastinal leakage, and free leakage into the pleural space.^[3,10] The first case would be classified as the most severe IED requiring surgical drainage, especially when the deep neck, mediastinal, and pleural spaces were also involved. The second case would be classified as the least severe IED. Therefore, endoscopic intramural drainage may be a viable alternative treatment procedure.^[6-8]

For suppurative IED, drainage procedures were a sure choice for infection control.^[4-9] The infection source could be removed mechanically, and the pathogen could be collected for culture to prescribe the most suitable antibiotics. Although no esophagectomy was required in both cases, the course of infection was prolonged in the latter case.

Suppurative IED is a very rare disease that has only been reported in a single case reports in the medical literature. Gaining from our experience of two consecutive cases managed surgically (the first case of IED with a mucosal perforation) versus conservatively (the second case of nonperforated IED), we present a successful management of the disease employing a minimally invasive surgical drainage procedure through neck combined with video-assisted thoracoscopic mediastinotomy and laparoscopic jejunostomy for the more severe IED with a mucosal perforation. For suppurative IED, drainage procedure may be more beneficial than antibiotic treatment alone if the surgical risk is acceptable. The severity of IED

could be taken into consideration when deciding on the choice of drainage procedure.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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