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Author(s)	Shichinohe, Toshiaki; Ebihara, Yuma; Murakami, Soichi et al.
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Case report

Septic shock-related acute esophageal necrosis and stenosis -three cases of acquired esophageal stenosis presenting a similar clinical course

Author: Toshiaki Shichinohe[1], Yuma Ebihara[1], Soichi Murakami[1], Yo Kurashima[1], Taro Kuramae[1], Motoshi Kanai[1], Joe Matsumoto[1], Takahiro Tsuchikawa[1], Masanobu Kusano[2], and Satoshi Hirano[1]

Affiliations of the authors:

1. Department of Gastroenterological Surgery II, Hokkaido University Graduate School of Medicine.
2. Department of Surgery, Otaru Kyokai Hospital

Corresponding author: Toshiaki Shichinohe

The address, e-mail address, telephone and fax numbers of the corresponding author:

West-7, North-15, Kitaku, Sapporo, Hokkaido, 060-8638, Japan

shichino@med.hokudai.ac.jp, Tel: +81-11-706-7714, FAX: +81-11-706-7158

Abstract

We present a case of acute esophageal necrosis (AEN) and two acquired esophageal stenosis cases which showed a similar clinical course after an episode of septic shock. Extensive stenosis of the distal esophagus developed in all cases, which were refractory to dilation therapy and required surgical intervention. The etiology of the latter two cases was deduced from the stenosis after septic shock-induced AEN. Since the diagnosis of AEN is based on its characteristic endoscopic findings which can only be confirmed at the onset of the disease, we therefore called these cases "septic shock-related esophageal stenosis" or abbreviated to "septic esophageal stenosis". Further study of similar cases is required for understanding the etiology and management of AEN and the relevant disorder which may cause esophageal stenosis.

Key words

Acute esophageal necrosis
Stenosis
Septic shock
Septic shock-related esophageal stenosis

Introduction

Acute esophageal necrosis (AEN) is a rare acquired esophageal disorder which may cause esophageal stenosis. Gurvits et al. reviewed 88 cases of AEN and reported that initial symptoms were typically gastrointestinal bleeding and cardiovascular events and shock [1]. Diagnosis is done by endoscopic finding with the confirmation of diffuse necrotic esophageal mucosa (so-called "black esophagus") predominantly affecting the distal esophagus. Successive occurrence of complications was observed in 42% cases of AEN, development of esophageal stricture or stenosis (10.2%), mediastinitis/abscess (5.7%), perforation (6.8%), and the overall mortality rate was 31.8% [1].

Since the diagnosis of AEN is based on the characteristic endoscopic appearance at the onset of the disease, it is assumed that some of AEN-caused stenosis cases are not being diagnosed because of a lack of endoscopic examination. And since the development of stricture takes weeks to months after the occurrence of AEN to appear, it would be difficult to associate stenosis development and past episode, and it would result in the diagnosis of "idiopathic esophageal stenosis".

In this report, we present a typical case of AEN and two such cases secondary to septic shock and propose the diagnostic name of "septic shock-related esophageal stenosis" or abbreviated to "septic esophageal stenosis"

Case report

Case 1: acute esophageal necrosis

A 76-year-old male with a medical history of ossification of posterior longitudinal ligament, neurogenic bladder, subdural hemorrhage, hypertension, and benign prostatic hyperplasia, developed septic shock caused by retroperitoneal abscess after a transurethral lithotomy operation for cystolithiasis, resection of urinary bladder adenoma, and cystostomy. Hematemesis developed accompanied with vital shock with leucocytosis requiring vasopressors. Emergent upper gastrointestinal fiberscope (UGF) revealed so-called "black esophagus" with the color change of the mucosa accompanied with edema in the entire part of the distal esophagus and the patient was diagnosed as AEN (Fig. 1). Four days after the onset, the patient recovered from the septic shock and UGF revealed wide-spread erosion and ulcer rather than "black esophagus" but no stricture was observed. On the 9th day, UGF confirmed the same findings and percutaneous endoscopic gastrostomy (PEG) was placed in preparation of the possibility of subsequent development of stenosis after AEN, and oral intake was started thereafter. On the 22nd day, UGF revealed esophageal stenosis and upper gastrointestinal series (UGI) confirmed diffuse stenosis of the middle to lower thoracic esophagus. Since PEG was placed after the diagnosis of AEN, the patient tolerated well repetitive endoscopic balloon dilation therapy (EBD) but it was not effective to resolve the stenosis (Fig. 2). Therefore, six months after the onset of the AEN he finally was operated on. Taking into consideration the poor condition of the patient, a bypass operation with gastric conduit was chosen. No complication was observed and the patient discharged on the 43rd post-operative day (POD) with sufficient oral intake.

Case 2: “septic shock-related esophageal stenosis”

A 70-year-old female with a medical history of pacemaker placement for atrioventricular block, drug eluting stent placement for angina pectoris, bronchial asthma, hypertension, and deep vein thrombosis, developed acute promyelocytic leukemia (APL). She was treated with consolidation chemotherapy by idarubicin after differentiation therapy by all-trans retinoic acid. During the consolidation chemotherapy, septic shock developed caused by idarubicin-induced agranulocytosis. No hematemesis or melena was observed during the period. After recovery from septic shock, she was discharged from the hospital with complete remission of APL. Two months after the event of septic shock, she noticed swallowing difficulty, and then one month later she was admitted with dysphagia. UGF and UGI revealed stenosis of the middle to lower esophagus and a biopsy revealed granulation tissue without any overt malignancy (Fig. 3). Although the patient received repeated EBD with concomitant injection of steroid, the effects were transient. At the last trial of EBD, esophageal perforation occurred and emergent open thoracotomy was required for direct suture repair to the perforation; it was five months after confirmation of the stenosis. After recovering from the operation, she received a bypass operation with gastric conduit; it was seven months after confirmation of the stenosis. No postoperative complication was observed and the patient discharged 46 POD with sufficient oral intake.

Case 3: “septic shock-related esophageal stenosis”

A 53-year-old male with a medical history of perforation of sigmoid colon diverticulum two year before, repeated colonic perforation with the symptoms of acute generalized peritonitis. Sigmoidectomy and colostomy was performed as an emergency operation. Severe sepsis with systemic inflammatory response syndrome caused by perforative peritonitis and aspiration pneumonia developed after surgery. Tracheostomy and mechanical ventilation required for 39 days. No hematemesis or melena was observed during the period. Two months after the operation, he noticed swallowing difficulty. Subsequently to the diagnosis of segmental stricture of the lower thoracic esophagus by UGI and UGF, dilation therapy was performed three times (Fig. 4). Although the patient was discharged from the hospital with remission of dysphagia transiently, progressive stenosis recurred and an operation was required five months after the onset of colonic perforation. Ivor-Lewis esophagectomy was performed as a radical operation. The postoperative course was good and he was discharged 30 POD. Pathological finding of the resected specimen revealed sever fibrosis of the esophageal wall without any malignancy (Fig. 5).

Discussion

We present three cases of esophageal stenosis secondary to septic shock which required surgical intervention. One typical case of AEN and other two acquired esophageal stenosis cases represent very similar clinical features, except whether or not a symptom of hematemesis existed and observation by

UGF was performed (Table). The common clinical signs of these three cases were as follows; firstly, no pre-existing etiologies of esophageal disorder existed which may cause stricture such as peptic stricture, reflux, Achalasia, or chemical ingestion [3]; secondly, esophageal stenosis occurred secondly to septic shock; thirdly, the stenosis extended widely but limited within the lower esophagus; lastly, the stenosis developed gradually within several weeks' to several months after septic shock and conventional dilation therapies were ineffective (Table). Since the diagnosis of AEN is based on an endoscopic finding which can be observed at the initial period of the onset of the disease, we propose the symptom-based diagnostic name of "septic shock-related esophageal stenosis" or abbreviated to "septic esophageal stenosis" which defines esophageal stenosis which occurs subsequent to septic shock.

The most characteristic feature of AEN is extended esophageal stenosis but limited below the level of the bifurcation of the trachea and no involvement to the cardia of the stomach. Implicated etiology of AEN is a combination of tissue hypo perfusion mainly caused by shock-related microcirculatory dysfunction and impaired local defense barriers and corrosive injury from gastric contents [2]. The theoretical rationale that the lower part of the esophagus below the level of the bifurcation of the trachea is most susceptible to hypo perfusion is based on the following hypothesis: In contrast to the upper to middle esophagus that have sufficient blood supply from the branches of the bilateral inferior thyroid arteries and the bronchial arteries and vascular communications with the trachea, the blood supply of the lower esophagus is singly supported by few proper esophageal arteries.

The management of AEN is divided into two categories on the basis of acute and chronic phase; life-threating complications such as esophageal perforation, mediastinitis, and abscess occur in acute-phase which sometimes requires an emergency operation, in contrast, esophageal stenosis/stricture will develop in the chronic phase. Since about the half of AEN cases develop severe complications, preemptive placement of PEG, like our first case, will be helpful to support the nutrition during the treatments.

The management of AEN-induced esophageal stenosis is basically the same as that of peptic esophageal stricture which is treated by bougies or balloon dilation with antireflux therapy, steroid injection, and surgery [3]. Although there exist few reports of esophagectomy for AEN, one should not hesitate to operate in cases of persistent stricture which are refractory to conventional treatments such as repeated dilation therapy [4,5]. In the review literature, two cases out of 88 cases (2.3%) were operated for long stenosis middle-to-lower esophagus after AEN; one case was received three times EBD for six months, and the other case was received multiple dilation therapy by bougienage for four months [1,4,5]. In our cases, dilation therapies were tried for three to five months but the stricture was refractory to the treatment and one case developed perforation after frequent EBD for five months. According to the previous reports and our experiment, the case with the long esophageal stenosis and refractory to multiple EBD for several months is an indication of surgical intervention.

Different operative procedures were applied for our three cases; one was esophagectomy and the other

two were bypass operations. Since it is known that corrosive esophageal stenosis and esophageal achalasia may increase the risk of carcinogenesis presumably due to chronic inflammation, the choice of esophagectomy rather than bypass operation is prudent procedure [6]. On the other hand, a bypass operation will reduce the risk of an operation by avoiding open thoracotomy, so it is appropriate for patients in poor medical condition.

In conclusion, we experienced three cases of acquired esophageal stenosis presenting similar symptoms and clinical course secondary to septic shock. An additional number of similar cases is required for understanding the etiology and management of AEN and the relevant disorder which may cause esophageal stenosis.

Ethical Statement

This article does not contain any studies with human or animal subjects performed by any of the authors.

Conflict of interest

The authors declare that they have no conflict of interest.

References

1. Gurvits GE, Shapsis A, Lau N et al. Acute esophageal necrosis: a rare syndrome. J Gastroenterol. 2007; 42: 29-38.

2. Gurvits GE. Black esophagus: acute esophageal necrosis syndrome. World J Gastroenterol. 2010; 16: 3219-3225.

3. Spechler SJ. American gastroenterological association medical position statement on treatment of patients with dysphagia caused by benign disorders of the distal esophagus. Gastroenterology 1999; 117: 229-232.

4. Kim YH, Choi SY. Black esophagus with concomitant candidiasis developed after diabetic ketoacidosis. World J Gastroenterol. 2007; 13: 5662-5663.

5. Goldenberg SP, Wain SL, Marignani P. Acute necrotizing esophagitis. Gastroenterology 1990; 98: 493-496.

6. . Ribeiro U Jr, Posner MC, Safatle-Ribeiro AV, et al. Risk factors for squamous cell carcinoma of the oesophagus. Br J Surg. 1996; 83: 1174-1185.

Table

Summary of a case of acute esophageal necrosis and two cases of acquired esophageal stenosis

	Case 1	Case 2	Case 3
Characteristics of Pts.	76-yo. Male Post operation of the bladder	70-yo. Female Chemotherapy of leukemia	53-yo. Male Post operation of colonic perforation
Preexisting upper GI disorder	No		
Prodromal event	Septic shock Hematomesis	Septic shock	
Cause of septic shock	Retroperitoneal abscess	Agranulocytosis by chemotherapy	Peritonitis and pneumonia
Confirmation of AEN by UGF	Yes	No	
Stenotic region of esophagus	Lower esophagus, below the level of bifurcation of trachea		
Interval of septic shock to stenosis	3 week	2 month	2 month
Dilation therapy	Failed		
Interval of stenosis to operation	6 month	7 month	3 month
Operative procedure	Bypass		Esophagectomy

AEN; acute esophageal necrosis

UGF; upper gastrointestinal fiberscope

Figure legends

Figure 1.

Acute esophageal necrosis (case1)

Emergent upper gastrointestinal fiberscope revealed so-called “black esophagus” with color change of the mucosa accompanied with edema in the entire part of the distal esophagus (a). The lesion abruptly stopped at the gastroesophageal junction (b).

Figure 2.

Acute esophageal necrosis (case 1)

Esophageal stenosis was refractory to endoscopic balloon dilation therapy. UGI confirmed diffuse stenosis of the middle to lower thoracic esophagus (a). UGF revealed pinhole stenosis of the esophagus (b).

Figure 3.

“septic shock-related esophageal stenosis” (case 2)

Swallowing difficulty was noticed two months after the event of septic shock. Esophageal stenosis was confirmed afterward. UGI confirmed prolonged esophageal stenosis from the middle thoracic esophagus (a). UGF revealed pinhole stenosis of the esophagus (b).

Figure 4.

“septic shock-related esophageal stenosis” (case 3)

UGI and UGF revealed esophageal stenosis two months after the event of septic shock. Diffused esophageal stenosis from the middle thoracic esophagus was confirmed by UGI (a). UGF revealed pinhole stenosis of the esophagus (b).

Figure 5.

Microscopic features of resected specimen (case 3)

Esophageal wall was thickened to 8mm by the fibrosis. Esophageal epithelium thinning and predominant fibrosis of the esophagus spreading from the subepithelial layer to the part of muscularis propria were observed.

Atrophied lamina muscularis mucosae (arrow), esophageal mucous gland (arrow head) and dilated gland duct (bleak arrow). (H&E original magnification X10)

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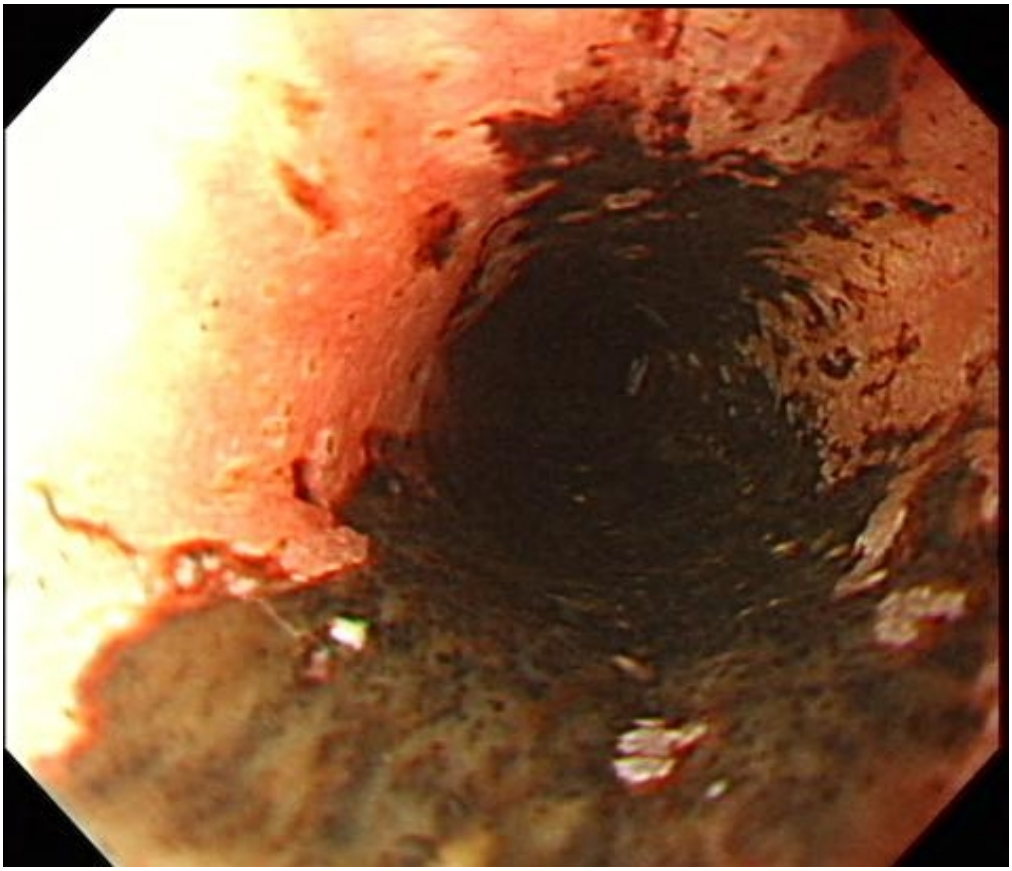
(fig.1a)



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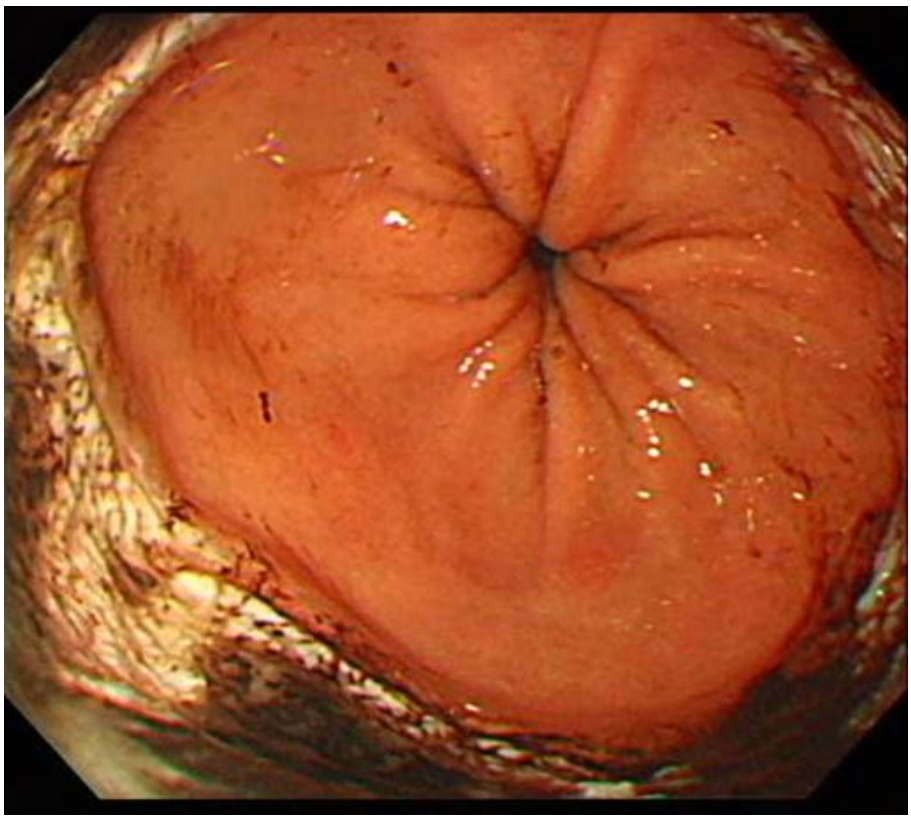
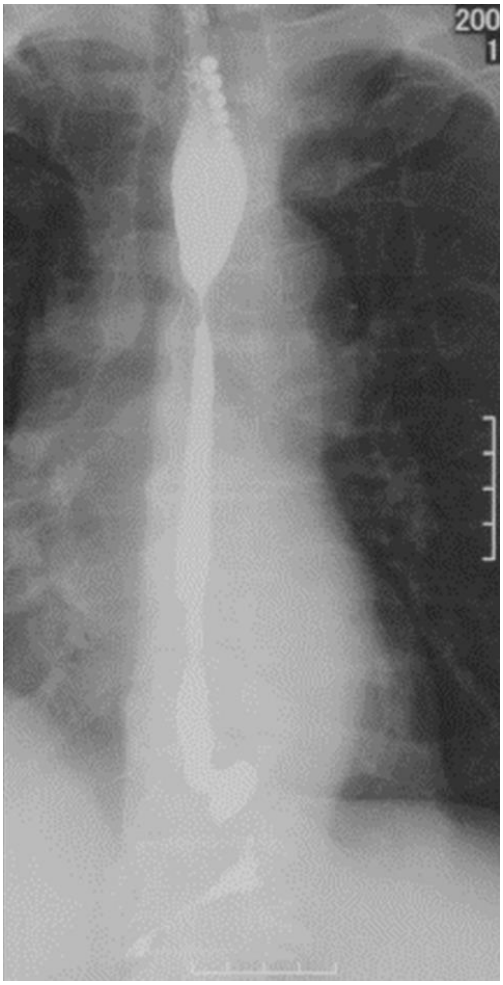


(fig.2a) 

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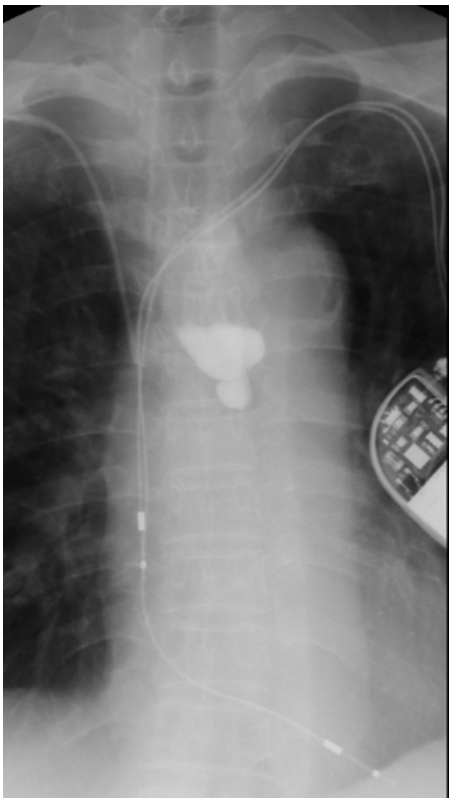
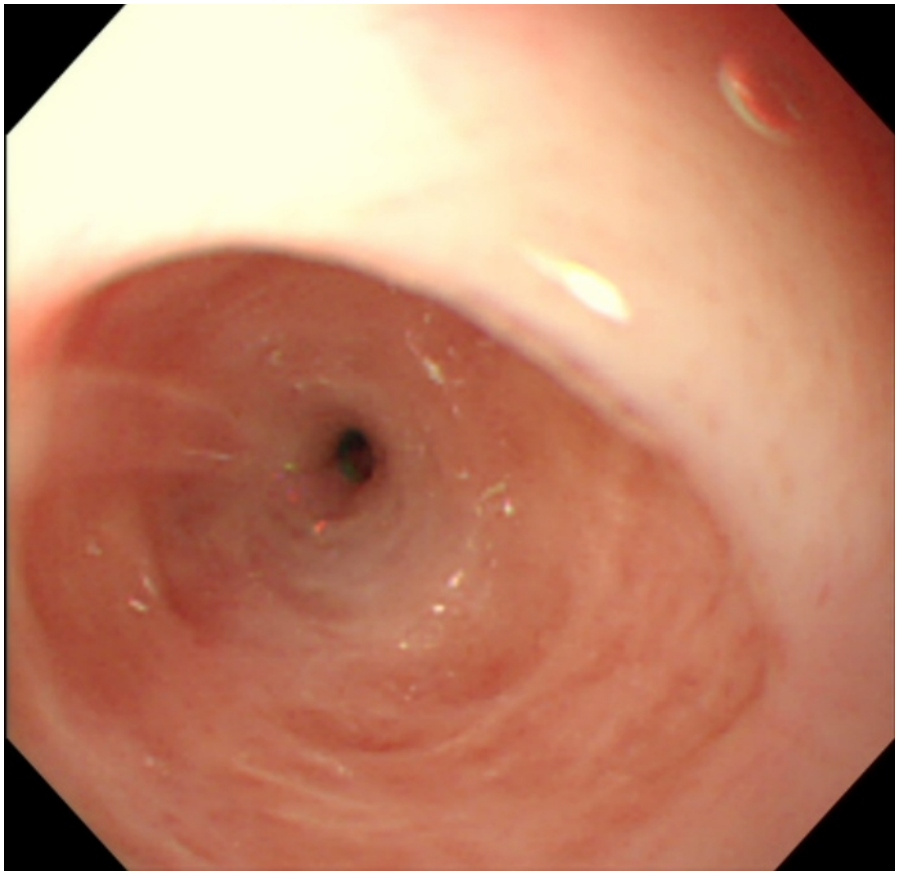
(fig.3a) 

(fig.3b) 

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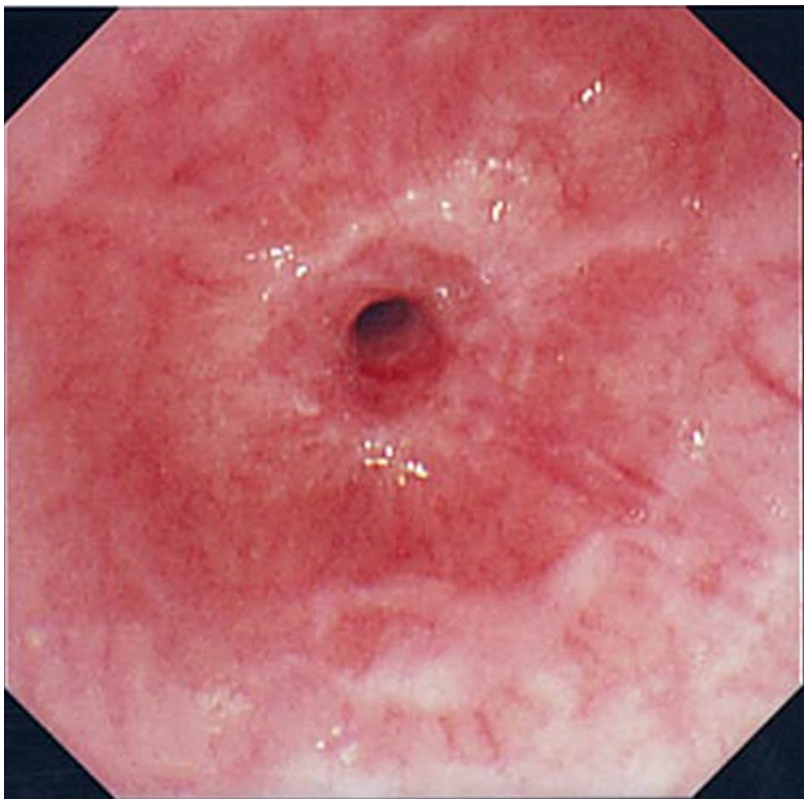

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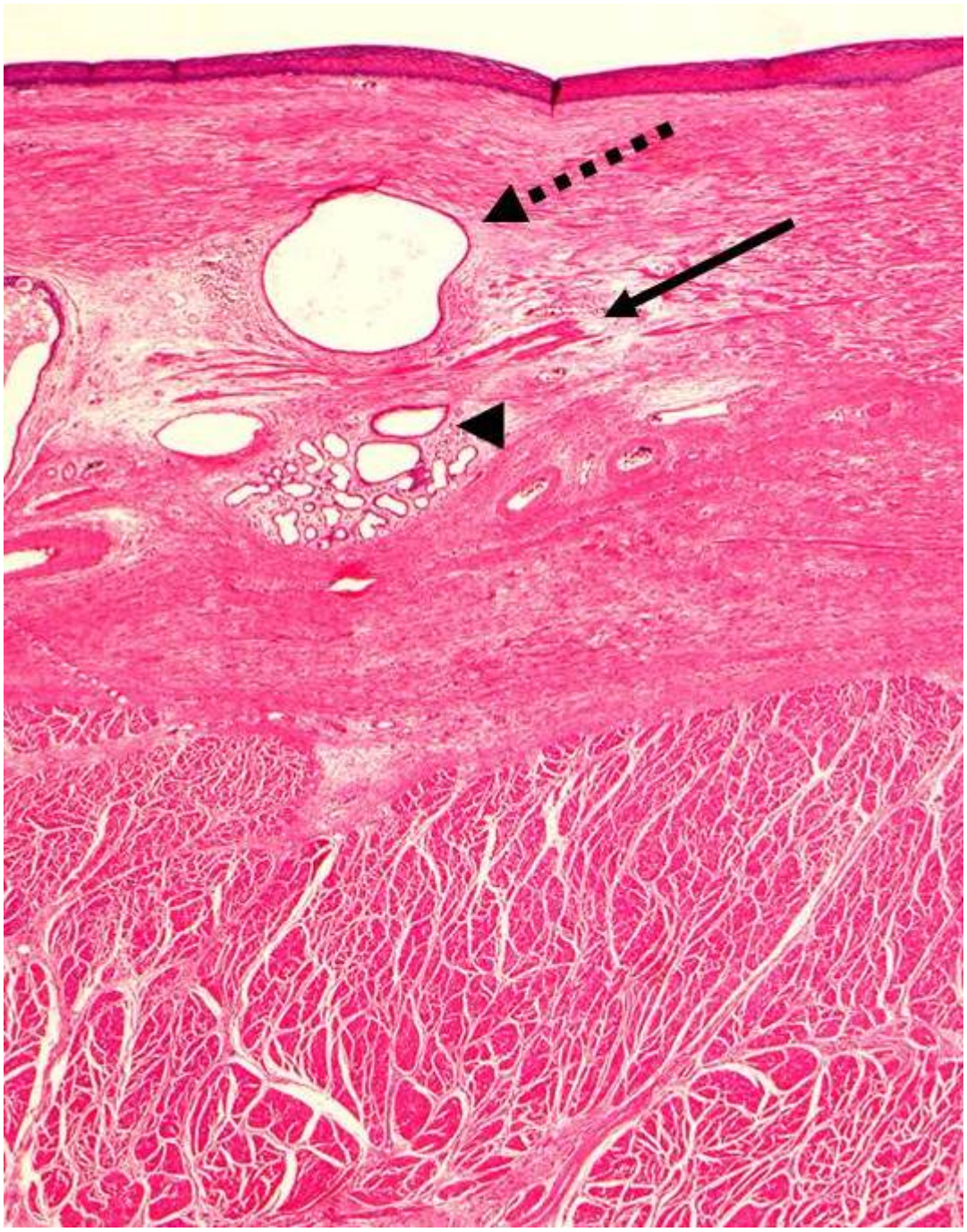
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