

Case Report

Acute Esophageal Necrosis as an Unusual Cause of Epigastric Pain in the Emergency Department

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Keywords

Esophageal diseases · Sepsis · Differential diagnosis

Abstract

Epigastric pain is a common complaint in the emergency department (ED). Clinicians require skills to differentiate the epigastric pain in the ED. Here, we report a case of acute esophageal necrosis (AEN) as a cause of epigastric pain in the ED. An 83-year-old woman with diabetes mellitus visited the ED because of worsening subacute epigastric pain, nausea, and anorexia. The patient's vital signs and general condition did not seem serious at first in the ED. Esophagogastroduodenoscopy revealed circumferential inflammation and necrosis of the esophageal mucosa. The patient was diagnosed with AEN and admitted. The patient's condition suddenly worsened on the sixth day. *Citrobacter koseri* was detected in blood culture, and although the patient was treated with antibiotics, she died on the twelfth day. In our case, epigastric pain, a common complaint in the ED in elderly women, was caused by AEN, an uncommon disease. The patient was seemingly stable at first but rapidly developed sepsis and died. In this case, we identified two important clinical issues: (1) AEN is an uncommon cause of epigastric pain in the ED, but it is worth considering. (2) Once AEN is diagnosed, the clinician should engage in further investigations such as esophageal and blood culture tests and close follow-up of the clinical course, even if patients' condition does not appear to be serious.

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Introduction

Abdominal pain is a chief complaint that clinicians frequently encounter in the emergency department (ED). Epigastric pain is common type of abdominal pain, accounting for 16.5% of all abdominal pain [1]. In patients with epigastric pain, clinicians should consider esophageal, gastrointestinal, hepatobiliary, and cardiovascular diseases [2]. Clinicians require skills to differentiate the common complaints in the ED.

Acute esophageal necrosis (AEN) is a rare disease. The main complaints include such as gastric bleeding and epigastric pain [3]. AEN is diagnosed using esophagogastroduodenoscopy (EGD), which shows diffuse circumferential black-colored distal esophageal mucosa [4]. Most patients with AEN have underlying medical conditions [4]. AEN is often triggered by serious illnesses such as diabetic ketoacidosis (DKA), dehydration, or shock [5].

This report describes AEN as an uncommon cause of epigastric pain in the ED. Moreover, this report describes that AEN may be an early warning of severe disease, even in the absence of critical abnormalities.

Case Report

An 83-year-old woman presented to the ED via ambulance with epigastric pain, nausea, and decreased appetite that had worsened over the past few days. Moreover, she had the following comorbidities: diabetes mellitus (DM), hypertension (HT), heart failure, and rheumatoid arthritis. She was regularly taking medications including furosemide, candesartan cilexetil, amlodipine besilate, sitagliptin phosphate hydrate, and buccillamine. Her consciousness was clear and abnormal vital signs were only HT: blood pressure, 176/84 mm Hg; pulse rate, 81 bpm; SpO₂ (under ambient air), 97%; and body temperature, 36.9°C. The lung sounds were normal. The abdomen was soft and flat, and signs of peritoneal irritation were negative.

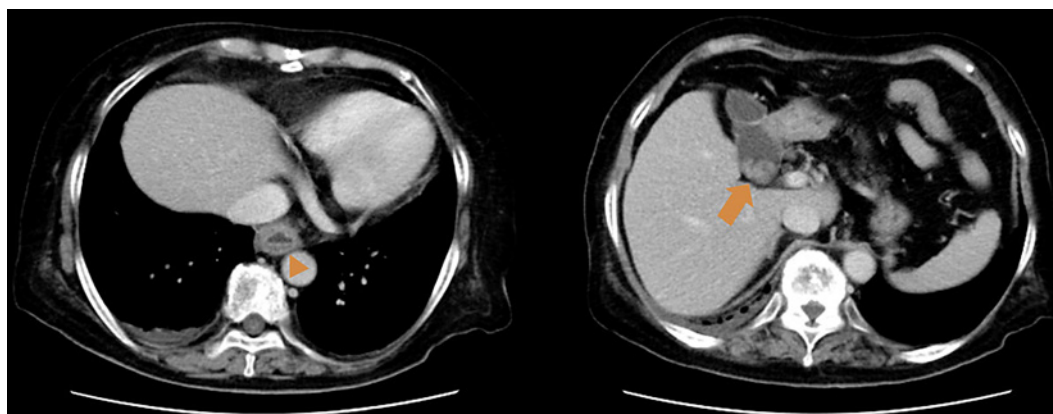
Laboratory tests showed an increased inflammatory response, hyperglycemia, and coagulation abnormalities (Table 1). Chest and abdominal radiographic findings did not explain the cause of the epigastric pain. Electrocardiography was suggestive of only hypertensive heart disease. Thoracoabdominal contrast-enhanced computed tomography (CT) showed thickened edema of the lower esophagus. Cholelithiasis was detected, but there was no evidence of cholecystitis (Fig. 1). Based on the CT findings, we determined that esophagitis was the cause of the symptoms.

The emergency physician referred the patient to a gastroenterologist for further investigation. EGD was performed on the first day, and circumferential inflammation and black changes in the esophageal mucosa were detected (Fig. 2). The patient was diagnosed with AEN and admitted to the general gastroenterology unit. Fasting was commenced, and vonoprazan, a proton pump inhibitor, was administered orally at 20 mg daily. Follow-up EGD on the sixth day showed improvement in the AEN finding. On the eighth day, the patient's condition rapidly worsened, resulting in a fever of 40°C. Other vital signs were as follows: blood pressure, 140/104 mm Hg; heart rate, 110 beats/min; and oxygen saturation, 96%. *Citrobacter koseri* was detected in the blood culture; however, the source of the bacteria was unknown. A urine test, contrast-enhanced CT, and abdominal ultrasonography revealed no specific organ findings; thus, pneumonia, urinary tract infection, and intra-abdominal infection were ruled out. No skin findings suggestive of peripheral venous route infection were admitted. The possibility of bacterial invasion from AEN lesions remained. Surgical therapy was not selected because there was no esophageal perforation or mediastinal abscess. Tazobactam/piperacillin was administered, which was sufficiently sensitive to *C. koseri*. However, the patient's condition deteriorated, and she died on the twelfth day.

Table 1. Laboratory findings on the ED admission

Hematology			Coagulation		
White blood cell count	11,300	/ μ L	PT-INR	1.1	
Red blood cell count	383×10^4	/ μ L	Activated partial thromboplastin time	25.1	sec
Hemoglobin	12	g/dL	Fibrinogen/fibrin degradation products	11.9	μ g/mL
Hematocrit	35.3	%	D-dimer	5	μ g/mL
Platelet	24.2×10^4	/ μ L	Myocardial marker		
Biochemistry			High sensitive troponin-I	6.6	pg/mL
Aspartate aminotransferase	17	IU/L	Creatinine kinase MB fraction	1.8	ng/mL
Alanine aminotransferase	6	IU/L	Blood gas analysis		
Lactate dehydrogenase	230	IU/L	pH	7.46	
Total bilirubin	1.1	mg/dL	pCO ₂	38.4	mm Hg
γ -Glutamyl transpeptidase	10	IU/L	HCO ₃ ⁻	26.9	mmol/L
Creatine kinase	94	IU/L	Anion gap	8.6	mmol/L
Glucose	232	mg/dL	Lactate	1.8	mg/dL
Hemoglobin A1c	6.2	%			
Total protein	7.2	g/dL			
Albumin	3.3	g/dL			
Blood urea nitrogen	14.6	mg/dL			
Creatinine	0.51	mg/dL			
Sodium	140	mEq/L			
Potassium	4.3	mEq/L			
Chloride	101	mEq/L			
C-reactive protein	4.5	mg/dL			

PT-INR, prothrombin time-international normalized ratio.


Fig. 1. Thoracoabdominal contrast-enhanced CT images showing edema on the esophagus (arrowhead) and cholelithiasis (arrow).

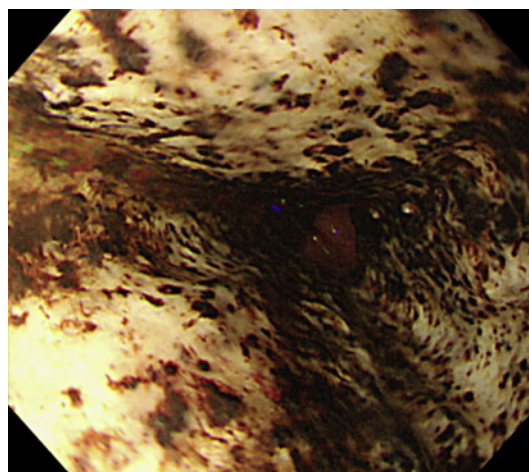


Fig. 2. EGD findings from the ED showing black changes on the esophageal mucosa.

Conclusion

In our case, epigastric pain, a common complaint in the ED in elderly women, was caused by AEN, an uncommon disease. The patient was seemingly stable and responded well to conservative treatment but rapidly developed sepsis and died. In this case, we identified two important clinical issues: (1) AEN is an uncommon cause of epigastric pain in the ED, but it is worth considering. (2) Once AEN is diagnosed, the clinician should engage in further investigations such as esophageal and blood culture tests and close follow-up of the clinical course, even if patients' condition does not appear to be serious.

First, AEN is a possible cause of epigastric pain that should be considered in the ED. Abdominal pain is a common complaint in the ED. 12.6% of adult patients in the general medical department of a university hospital experienced abdominal pain [1]. Epigastric pain is one of the common types of abdominal pain and accounts for 16.5% of abdominal pain [1]. Therefore, clinicians in the ED need to be skilled in the diagnosis of the causes of epigastric pain. The common differential diagnoses for epigastric pain include gastroduodenal ulcer, cholecystitis, and acute coronary syndrome [2]. In this case, the epigastric pain was caused by AEN. Although AEN is rare, it is a disease that clinicians may encounter in the ED and should be aware of. AEN presents with characteristic black changes in the esophageal mucosa on EGD [4] and was first reported by Goldenberg et al. in 1990 [6]. AEN has multiple causes, including ischemic damage to the esophagus, reflux of gastric juice leading to further damage, and an inadequate mucosal immune response [7]. The main complaints of AEN are gastrointestinal bleeding, chest and epigastric pain, nausea, vomiting, and anorexia [3]. Risk factors include male sex, older age, trauma, and paraesophageal hernia [7]. Comorbidities that increase the risk of developing AEN include DM, DKA, HT, cardiovascular disease, atherosclerosis, chronic kidney disease, and chronic liver disease [7]. Moreover, 83% of cases have underlying diseases [8]. This case suggests the importance of performing EGD to confirm the diagnosis of AEN when a patient complains epigastric pain with a thickened distal esophagus on CT. In addition to distal esophagus thickness, hiatal hernia, distended fluid-filled stomach, and external compression of the esophagus by a large mediastinal process were also reported as CT findings in AEN [8].

Second, AEN is potentially associated with serious disease states; hence, clinicians should investigate carefully. AEN is reported to have a poor prognosis, with a mortality rate of 31.8%; however, in only few cases does AEN itself cause death, and many cases improve with conservative treatment if there are no complications [4]. Nonetheless, the development of AEN is

correlated with the risk of mortality from underlying severe diseases, such as DKA, dehydration, and shock, and should be considered a factor in poor prognosis [5, 9]. Surgical intervention for AEN is indicated in cases of perforation of the esophagus with mediastinitis or abscess formation [7]. In our case, the patient did not appear to be seriously ill initially, but she developed sepsis caused by *C. koseri* and died in a short time. *C. koseri* is an enteric bacterium that often causes infections in immunosuppressed patients. Drelichman et al. [10] reviewed 31 cases of *Citrobacter* spp. bacteremia and reported that the most common entry points were the urinary tract (39%), gastrointestinal tract (27%), and wound (10%) and mortality rate was 48%. Our patient had DM and was immunosuppressed. Our investigation revealed no evidence of an infectious focus for *C. koseri*. Since the infection was not expected at the time of admission, blood cultures were not collected until the patient's medical condition worsened. Therefore, it is impossible to determine whether *C. koseri* infection was the cause or a complication of AEN as both are plausible. We speculate on the possibility of bacterial invasion from AEN lesions. Esophageal biopsy and culture at initial EGD may have detected local infection before the general condition deteriorated. In a previous report reviewing 112 cases of AEN, bacteria were detected in one esophageal biopsy [4]. We endorse the proactive approach of esophageal biopsy in AEN.

In conclusion, patients with epigastric pain in the ED should be assumed to have AEN. The onset of AEN itself may be a sign of a critical situation caused by background diseases or subsequent complications. We suggest a detailed investigation including esophageal and blood culture and careful follow-up, even for seemingly stable patients with AEN.

Statement of Ethics

Ethical approval is not required for this study in accordance with local or national guidelines. Since case reports are not included in the scope covered by the Ethical Guidelines for Medical and Biological Research Involving Human Subjects issued by the Ministry of Health, Labour and Welfare (MHLW), it is the general interpretation in Japan that case reports are published without Ethics Committee review. And written informed consent was obtained from the participant's daughter for publication of the details of their medical case and any accompanying images. The patient was deceased and therefore incapable of consenting.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Tomoki Ito designed the report and wrote the initial draft of the manuscript. He approved the final manuscript. Kasumi Satoh substantially contributed to the conception of the report and critically revised the manuscript. She approved the final manuscript. Kotaro Sakaki and

Masaru Sakusabe substantially contributed to the collection of clinical information and critically revised the manuscript. They approved the final manuscript.

Data Availability Statement

All data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.

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