Black esophagus: A case of acute esophageal necrosis associated with diabetic ketoacidosis

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April 6, 2021

Abstract

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Abstract

Acute esophageal necrosis (AEN), commonly referred to as "black esophagus", is a rare clinical disease. Though AEN remains a rare cause of upper gastrointestinal bleeding, it is a potentially life-threatening condition. We present a case of AEN associated with diabetic ketoacidosis.

Key Clinical Message

A clinician should be aware of acute esophageal necrosis as a potential cause of upper gastrointestinal bleeding when faced with patients with diabetic ketoacidosis.

KEY WORDS

acute esophageal necrosis, black esophagus, acute necrotizing esophagitis, diabetic ketoacidosis

1 INTRODUCTION

Acute esophageal necrosis (AEN), commonly referred to as "black esophagus" or "acute necrotizing esophagitis", is a rare cause of upper gastrointestinal bleeding. It was first described in the medical literature in 1990 by Goldenberg et al., and was brought into the spotlight in 2007 by Gurvits et al. (1,2). In endoscopic examinations, the prevalence of AEN is rare and reported to be up to 0.28% (3,4). The precise etiology of the disease remains unknown, but is presumed to be multifactorial with mucosal ischemia, injury by the reflux of gastric chemical contents, and an impaired mucosal barrier associated with debilitated physical states (3,4). It has been reported that patients usually present multiple comorbidities, the most common being diabetes mellitus, followed by hypertension, alcohol abuse, chronic kidney disease, coronary heart disease, dyslipidemia, peripheral artery disease, malignancy, liver cirrhosis, gastroesophageal reflux disease, congestive heart failure, and chronic obstructive pulmonary disease (5). Diabetes mellitus leads to marked microvascular disease that can be an important contributor to the development of esophageal necrosis (3,6-8).

Clinical presentation is remarkable for upper gastrointestinal bleeding, with most patients presenting with hematemesis or melena (3,4). Serious acute complications include perforation and mediastinitis (9). The most common long-term complication is stricture formation, which may occur in up to 10% of cases (3,9). A mortality rate as high as 32% has been reported, likely related to comorbidities (3,4). The mortality rate specific to AEN is approximately 6%, largely due to esophageal perforation (2-4,9). A standard treatment for AEN is not yet established, but most reports have recommended the treatment of coexisting clinical conditions, systemic resuscitation with intravenous fluid therapy, glycemic control, use of aggressive intravenous proton pump inhibitors, and parenteral nutrition (3,4,9).

Here, we report a case of "black esophagus" induced by diabetic ketoacidosis (DKA).

2 CASE REPROT

A 66-year-old male presented to our hospital with coffee-ground emesis, dyspnea, and general malaise. He also had abdominal pain and nausea. His medical history included type 2 diabetes mellitus, obstructive pulmonary disease, and alcohol abuse. He had not been taking his medication, including insulin, for several days due to nausea. His vital signs were as follows: Glasgow Coma Scale, 15 (E4V5M6); body temperature, 35.5 ; blood pressure, 103/79 mmHg; pulse, 100 beats per minute; and percutaneous oxygen saturation with a flow rate of 2 liters per minute through a nasal cannula, 100%. A physical examination revealed pallor conjunctiva and a coffee ground-like substance around the mouth. The abdomen was mildly tender without guarding. Other physical examinations revealed no remarkable findings. Laboratory analysis revealed severe hyperglycemia (730 mg/dL), normocytic anemia (hemoglobin level, 7.7 g/dL; mean corpuscular volume, 100.4 fL), high serum potassium (7.6 mEq/L), and a high level of blood urea (98.7mg/dL). A urine sample was positive for ketones and glucose, and serum β-hydroxybutyrate was elevated (2,132 μmol/L). Arterial blood gas analysis showed metabolic acidosis (pH, 7.29; HCO_3, 10.5 mmol/L) (Table 1). Collectively, the patient was diagnosed with DKA and upper gastrointestinal bleeding.

The patient was resuscitated with intravenous fluids, and administered insulin by intravenous infusion for treatment of DKA. After approximately 48 h of insulin infusion, the daily plasma glucose profile improved. The patient underwent esophagogastroduodenoscopy (EGD) which showed a circumferential necrosis of the middle and distal portions of the esophagus with an abrupt transition at the gastroesophageal junction (Figure. 1A, 1B). A chest computed tomography revealed a thickened distal esophagus but excluded the presence of esophageal perforation. EGD also revealed a gastric ulcer on the lesser curvature of the upper body of the stomach with an exposed blood vessel (Figure. 1C). The exposed blood vessel was cauterized with hemostatic forceps. From the endoscopic findings, acute necrotizing esophagitis and hemorrhagic gastric ulcer were diagnosed. The patient was then treated with an intravenous proton pump inhibitor. A second EGD, performed one day after admission, showed no remarkable findings with circumferential black discoloration at the middle and distal portion of esophagus as compared to those the day before (Figure. 2A, 2B), and hemostasis of the gastric ulcer. The patient continued to improve with conservative management and he was subsequently discharged from the hospital in a stable condition. An EGD repeated 14 days after

discharge showed complete healing of the necrotic-appearing mucosal change without stricture formation of the esophagus (Figure. 3A, 3B).

3 DISCUSSION

Black esophagus, also known as ANE, was first described by Goldenberg et al. in 1990 (1), and gained prominence after a series of cases were reported in 2007 (2). AEN has remained a rare diagnosis with an estimated prevalence of up to 0.28% (3,4). It has an endoscopic finding characterized by diffuse, circumferential black discoloration of the mucosa that affects the distal part of the esophagus and stops abruptly at the gastroesophageal junction (1-4,7,10).

Though the exact etiology of the disease remains unclear, it is presumed to be multifactorial with ischemia, impaired intrinsic mucosal barriers, and massive reflux of gastric contents that acutely exceeds the protective and regenerative properties of vulnerable esophageal mucosa enabling both ischemic and chemical injury of the esophagus (3,4,8,11-13). Chronic predisposing conditions and poor nutritional status may also contribute to decreased mucosal buffering and may potentiate the mechanisms by which esophageal necrosis is progressed.

Diagnosis of AEN is based on endoscopic findings which consist of a circumferentially black-appearing esophageal mucosa that abruptly ends at the gastroesophageal junction and transitions to normal gastric mucosa (1-4,7,10). Biopsy is supportive and obtained to exclude other etiologies, but is not required to establish the diagnosis (3,4). Patients are typically older males with multiple medical comorbidities, which increase the likelihood of AEN (2-4). Demographically, hyperglycemia is seen in almost 90% of patients with this condition (14,15). Moreover, disease associations include vascular compromise, hypertension, chronic kidney disease, malignancy, malnourishment, gastric outlet obstruction, and in this case, diabetes mellitus and alcohol abuse (3,4). The clinical presentation is remarkable for upper gastrointestinal bleeding, with most patients presenting with hematemesis or melena, comparable to our case (3,4). Other symptoms may include abdominal pain, nausea, vomiting, dysphagia, fever, and syncope (3,4). The patient presented with abdominal pain and nausea. Laboratory findings are nonspecific, but it has been reported that associated laboratory abnormalities may include anemia, leukocytosis, and hyperlactatemia (3,4), all of which are comparable to the patient.

There is no specific therapy for AEN. Management of AEN should be directed at correcting the underlying medical condition and protective care with hemodynamic resuscitation, total parenteral nutrition, and the administration of antacid therapy with aggressive intravenous proton pump inhibitors (3,4). Enteral feeding is not recommended because of the risk of perforation unless there is persistent vomiting (3,4,9). Moreover, prophylactic antibiotics are not necessary. The decision to initiate antimicrobial therapy should be individualized based on clinical and objective findings of serious infection (3,4,9,16). The patient presented no sign of infection. The most common long-term sequela of AEN is stricture formation, which can complicate up to 10% of all cases (3,4,9). No stricture formation was shown in our case on follow-up EGD.

The correlation between DKA and AEN has been described in the literature (14,17-23). DKA has been reported to be one of the most common triggering events for AEN (3,4,9). Although the precise mechanism remains unknown, several pathogenic mechanisms may possibly be implicated. The presence of long-standing diabetes mellitus predisposes the patient to the development of atherosclerosis leading to an increased risk of ischemia (3,4,14,19,24,25). It has been suggested that poor nutritional status with hemodynamic instability, and hyperglycemia in DKA can lead to poor vascular flow and an impaired mucosal barrier from corrosive injury of gastric contents (3,4,8,14,19,24,25). Moreover, gastric stasis, which can occur in the absence of neuropathy during DAK, can induce gastroesophageal reflux by which esophageal necrosis is accelerated (3,4,19). DKA can also result in profound osmotic diuresis and fluid loss leading to hypoperfusion of the distal esophagus (3,4,14,18).

In summary, we presented a case of AEN complicating diabetic ketoacidosis. Though AEN remains a rare cause of upper gastrointestinal bleeding, it is a potentially life-threatening condition. Notably, up to 9% of patients with DKA have evidence of upper gastrointestinal bleeding, but less than one-third of those

undergo endoscopy to determine the underlying cause (10). Therefore, a clinician should be aware of AEN as a potential cause of upper gastrointestinal bleeding when faced with an elderly, possibly poorly controlled diabetic patient or when additional significant comorbidities are present.

CONFLICT OF INTERESTS

The authors declare that they have no competing interests.

AUTHORS' CONTRIBUTIONS

DK and AN: wrote the manuscript. KS, YO, KN, YN, and HI: contributed to the drafting.

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TABLE AND FIGURE LEGENDS

TABLE 1. Laboratory data on admission.

FIGURE 1. Esophagogastroduodenoscopy on admission showed a circumferential, diffusely necrotic mucosa necrosis from the middle (A) to distal portions (B) of the esophagus with an abrupt transition at the gastroesophageal junction. (C) Gastric ulcer on the lesser curvature of the upper body of the stomach with an exposed blood vessel.

FIGURE 2. Images from a second esophagogastroduodenoscopy performed one day after admission. A circumferential black discoloration of mucosa from the middle (A) to the distal portion of the esophagus (B).

FIGURE 3. Esophagogastroduodenoscopy performed 14 days after discharge. A complete healing of the necrotic-appearing mucosal changes from the middle (A) to the distal portion (B) of the esophagus.

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`TABLE 1.pdf` available at https://authorea.com/users/405958/articles/516795-black-esophagus-a-case-of-acute-esophageal-necrosis-associated-with-diabetic-ketoacidosis

FIGURE 1.

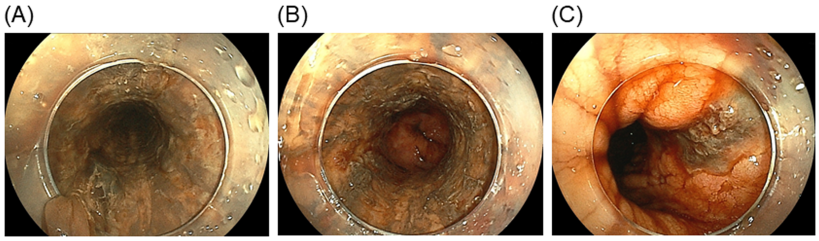

FIGURE 2.

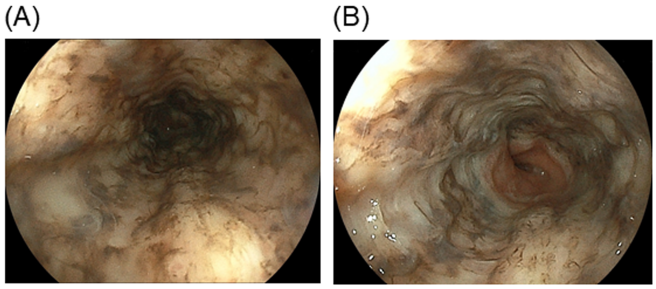

FIGURE 3.