

Acute esophageal necrosis after cellulitis in an obese patient with diabetes mellitus

Sho Tanaka¹ , Midori Fujishiro^{2*}, Ryoji Ichijima³, Genta Kohno², Masanori Abe¹, Hisamitsu Ishihara²

¹Division of Nephrology, Hypertension and Endocrinology, ²Division of Diabetes and Metabolic Diseases, and ³Division of Gastroenterology and Hepatology, Department of Internal Medicine, Nihon University School of Medicine, Tokyo, Japan

Keywords

Diabetes mellitus, Endoscopy, Esophagitis

*Correspondence

Midori Fujishiro
 Tel.: +81-3-3972-8111
 Fax: +81-3-3972-8199
 E-mail address:
 fujishiro.midori@nihon-u.ac.jp

J Diabetes Investig 2020; 11: 250–252

doi: 10.1111/jdi.13104

ABSTRACT

A 59-year-old obese Japanese man with poorly controlled type 2 diabetes mellitus presented with severe heartburn for 3 days after inguinal cellulitis and exacerbated glycemic control, without any signs of upper gastrointestinal bleeding. The patient had a high plasma glucose level (34.0 mmol/L) and was dehydrated. Emergent esophagogastroduodenoscopy showed black discoloration predominantly affecting the lower esophagus; thus, acute esophageal necrosis (AEN) was diagnosed. This black discoloration was not present on esophagogastroduodenoscopy 20 days prior to presentation, and disappeared 6 days after conservative treatment. To conclude, acute esophageal necrosis should be considered if a patient in marked hyperglycemic status presents with unendurable heartburn, even when upper gastrointestinal bleeding is not observed or recent esophagogastroduodenoscopy was unremarkable.

INTRODUCTION

Exacerbated glycemic profiles after infectious disease in patients with diabetes mellitus are frequently encountered in clinical practice. Marked hyperglycemia sometimes dehydrates patients through osmotic diuresis, and gives rise to renal insufficiency. Acute esophageal necrosis (AEN) is a rare but potentially life-threatening syndrome that preferentially affects dehydrated elderly men with hyperglycemia, and arises together with upper gastrointestinal bleeding in almost all such patients¹. Herein, we report a case of AEN without any initial signs of upper gastrointestinal bleeding in whom subsequent esophagogastroduodenoscopy (EGD) led to a diagnosis of AEN.

CASE REPORT

A 59-year-old Japanese man with type 2 diabetes mellitus presented with spreading erythema in the inguinal region, followed by a deteriorated glycemic profile for 1 week, and heartburn for 3 days. His diabetes mellitus was unsatisfactorily controlled: the glycated hemoglobin level was 9.0%, despite 2,000 mg/day metformin, 4–6 units of insulin glulisine before meals and 20 units of insulin glargine at bedtime. The severe heartburn prevented the patient from eating solid meals, although a small amount of a liquid meal with suspended metformin and insulin was taken, and an accelerated deterioration in glycemic control

was subsequently noted. Episodes of melena, hematemesis or coffee-ground emesis were not apparent.

The patient was obese (body mass index 33.6 kg/m²), normotensive but tachycardic, and had a low-grade fever and dry mucous membranes. Right inguinal cellulitis with surface necrotic tissue complicated by a subcutaneous abscess (Figure 1) was observed. Laboratory analysis revealed a white blood cell count of 17,100/μL, blood urea nitrogen at 37.7 mg/dL, creatinine at 0.98 mg/dL, an estimated glomerular filtration rate of 61.5 mL/min/1.73 m² (a drop from a baseline level of 84.9 mL/min/1.73 m²), C-reactive protein at 25.4 mg/dL, plasma glucose at 34.0 mmol/L and a glycated hemoglobin level of 10.1%. Diabetic ketosis without acidosis was also observed.

Esophagogastroduodenoscopy during a medical checkup 20 days before presentation had shown only short-segment Barrett's esophagus (Figure 2a), whereas emergent EGD on arrival showed diffuse erosive esophagitis with black discoloration predominantly affecting the lower esophagus and abruptly interrupted at the gastroesophageal junction (Figure 2b); thus, a diagnosis of AEN was made.

The patient was managed by oral intake restriction, fluid replacement with isotonic solution, intravenous omeprazole 20 mg twice daily, intravenous insulin infusion, piperacillin-tazobactam 4.5 g three times daily, and by incision and drainage of the abscess. Prerenal azotemia, decreased estimated glomerular filtration rate and ketosis swiftly improved, with 8–10 mmol/L glucose achieved on hospitalization day 5. Follow-up EGD on day 6 showed a marked improvement of AEN

Received 8 April 2019; revised 6 June 2019; accepted 16 June 2019



Figure 1 | Cellulitis with a subcutaneous abscess in the lower inguinal region.

(Figure 2c). Thus, the intravenous omeprazole was switched to vonoprazan 20 mg orally once daily, and the intravenous insulin was switched to multiple daily injections in accordance with a restarted oral intake. The cellulitis healed by hospitalization day 16, and the patient was discharged.

Written informed consent was obtained from the patient for publication of this report. Formal ethics approval was waived, because this is a case report.

DISCUSSION

AEN, a syndrome endoscopically characterized by a diffuse black discoloration affecting the distal esophagus, was first described by Goldenberg *et al.*² in 1990. Although previous endoscopy series have described AEN as being very rare, with an estimated prevalence rate of between 0.01 and 0.28% of endoscopies, early diagnosis and management are required due to possible esophageal perforation and high mortality rates³.

Indeed, the overall mortality of patients with AEN was found to be 31.8%⁴. AEN frequently occurs in elderly persons and has a clear male predominance; nearly 90% of cases manifest melena, hematemesis or coffee-ground emesis^{1,4}. Hyperglycemia and diabetes mellitus are recognized risk factors for AEN because of a frequent co-existence; AEN with diabetic ketoacidosis has also sometimes been reported^{1,4-7}.

The most plausible explanations for the etiology of this case include gastroesophageal acid reflux, weakened mucosal defense and hypoperfusion, as these are associated with the developmental mechanisms of AEN⁴. Pre-existing Barrett’s esophagus indicates long-standing gastroesophageal reflux. Critical illness (severe cellulitis, after marked hyperglycemia and renal insufficiency in the present case) leads to diminished acid buffering, resulting in a weakened esophageal defense³. Furthermore, hemodynamic instability related to severe infection and dehydration due to hyperglycemic osmotic diuresis, suspected based on a physical assessment and laboratory findings, can involve rapid injury to esophageal tissue³.

The pitfalls of AEN management are highlighted in the present case. First is the lack of any signs of upper gastrointestinal bleeding. Because 7% of cases are accompanied by the acute life-threatening complication of esophageal perforation, which requires life-saving surgical intervention, the early recognition and management of AEN is vital^{1,4}. However, laboratory findings of AEN are not specific and largely due to the underlying disease^{1,4}. Therefore, clinicians carry out EGD when patients with multiple risk factors of AEN complain of heartburn, even in the absence of signs of upper gastrointestinal bleeding. Second is the dramatic difference between EGD findings on admission and the latest, coincidentally obtained, pre-morbid EGD findings. The development of AEN is thought to be rapid, having occurred within 18 h in one case⁸. This present case highlights how AEN can develop when previously trivial endoscopic findings were yielded, thus suggesting that a recent EGD showing unremarkable findings cannot rule out AEN in clinical practice.

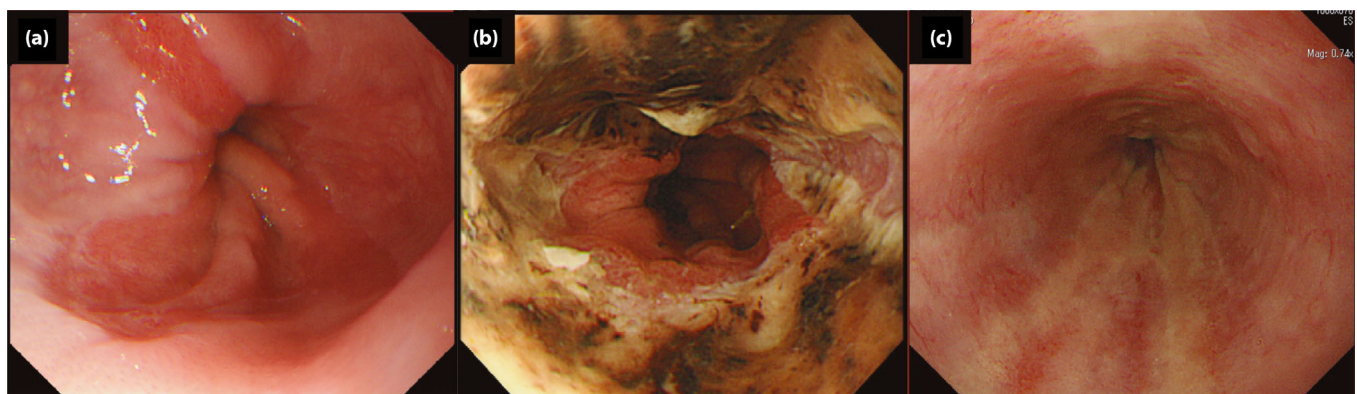


Figure 2 | Esophagogastroduodenoscopy findings. (a) Twenty days before presentation. (b) On admission. (c) On hospitalization day 6.

The deterioration in glycemic profile after preceding infectious disease in patients with poorly controlled diabetes mellitus is frequently encountered in a clinical setup. Such patients are sometimes dehydrated and thus at high risk of AEN. Clinicians should consider AEN and carry out timely EGD if a diabetes patient with a severely elevated glucose level complains of unendurable heartburn, even when signs of upper gastrointestinal bleeding are absent or a recent EGD was unremarkable.

DISCLOSURE

The authors declare 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. Goldenberg SP, Wain SL, Marignani P. Acute necrotizing esophagitis. *Gastroenterology* 1990; 98: 493–496.
3. Gurvits GE. Black esophagus: acute esophageal necrosis syndrome. *World J Gastroenterol* 2010; 16: 3219–3225.
4. Gurvits GE, Cherian K, Shami MN, *et al.* Black esophagus: new insights and multicenter international experience in 2014. *Dig Dis Sci* 2015; 60: 444–453.
5. Kim YH, Choi SY. Black esophagus with concomitant candidiasis developed after diabetic ketoacidosis. *World J Gastroenterol* 2007; 13: 5662–5663.
6. Haghbayan H, Sarker AK, Coomes EA. Black esophagus: acute esophageal necrosis complicating diabetic ketoacidosis. *CMAJ* 2018; 190: E1049.
7. Rigolon R, Fossa I, Rodella L, *et al.* Black esophagus syndrome associated with diabetic ketoacidosis. *World J Clin Cases* 2016; 4: 56–59.
8. Marie-Christine BI, Pascal B, Jean-Pierre P. Acute necrotizing esophagitis: another case. *Gastroenterology* 1991; 101: 281–282.