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Case Report

Gastric Mucormycosis in an Alcoholic with Review of the Literature

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Abstract

Mucormycosis is a rare disease with a high mortality rate. Common sites of mucormycosis include cerebral, pulmonary, gastrointestinal and disseminated lesions. We present the case of a 58-year-old woman with chronic alcoholism who was admitted to our intensive care unit with the initial presentation of abdominal pain. Colonic perforation was diagnosed upon surgical intervention. The patient soon developed upper gastrointestinal bleeding, and two masses at the body and antrum of the stomach were found on endoscopy. Pathological examination results showed invasive mucormycosis. The association of alcoholism and mucormycosis is emphasized in this case. (*Tzu Chi Med J* 2007;19(3):169–172)

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1. Introduction

Mucormycosis is a common name given to several different diseases caused by fungi of the order Mucorales (1). It is commonly involved in rhinocerebral infections in patients with diabetes or malignancy, as well as pulmonary and disseminated manifestations in patients with hematological malignancies. The gastrointestinal form of the disease is primarily found in patients suffering from extreme malnutrition (2). Here, we present a rare case of gastric mucormycosis in a patient with schizophrenia and chronic alcoholism.

2. Case report

A 58-year-old woman with schizophrenia was admitted to the emergency department complaining of persistent severe abdominal pain and cold sweats for 1 hour. Her medical history was significant for chronic alcoholism, hypertension with irregular control of medication, impaired glucose tolerance without oral medication, rectal polyp status post local excision 10 months prior to this admission, and chronic constipation for 4 years requiring manual discompaction.

Upon arrival at the emergency department, the patient's blood pressure was 206/112 mmHg. Abdominal examination revealed diffuse tenderness with hypoactive bowel sounds. Furthermore, laboratory study results showed hyponatremia (Na, 123 mmol/L) and hypokalemia (K, 2.7 mmol/L). An upright chest X-ray showed subphrenic free air, which suggested bowel perforation.

The surgeon performed an emergency laparotomy and found multiple perforations in the ascending colon with feces spreading within the entire abdominal

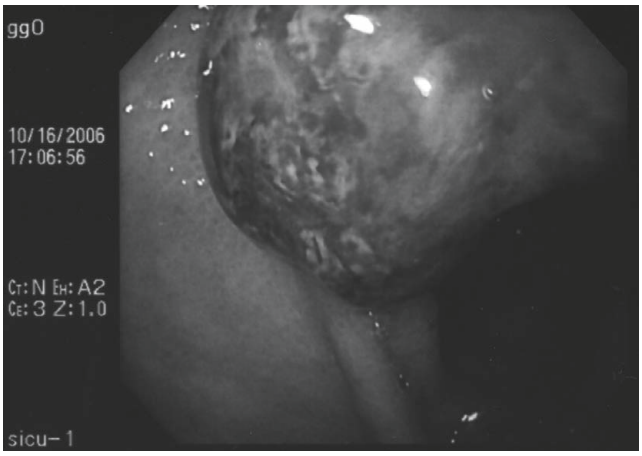

Fig. 1 — Endoscopy reveals a 4-cm mass in the gastric antrum. Some A2 ulcers can be seen on the surface of the masses.

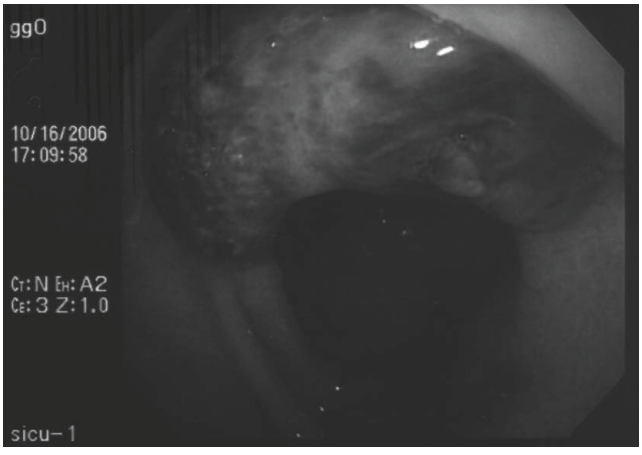

Fig. 2 — Endoscopy reveals a 5-cm mass on the posterior wall of the gastric body.

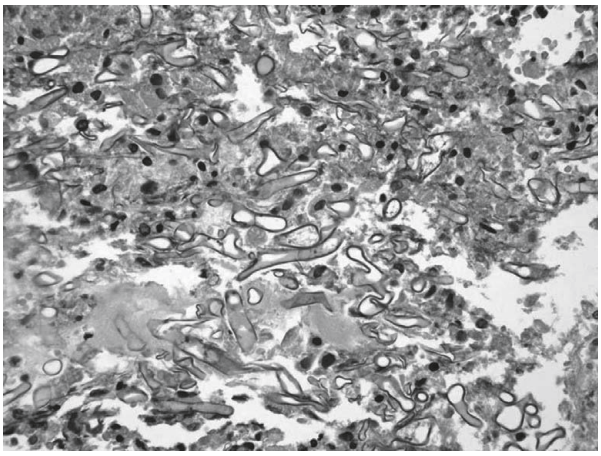

Fig. 3 — Photomicrograph shows gastric necrotic mucosal surface with numerous hyphae (hematoxylin & eosin; original magnification, 400×).

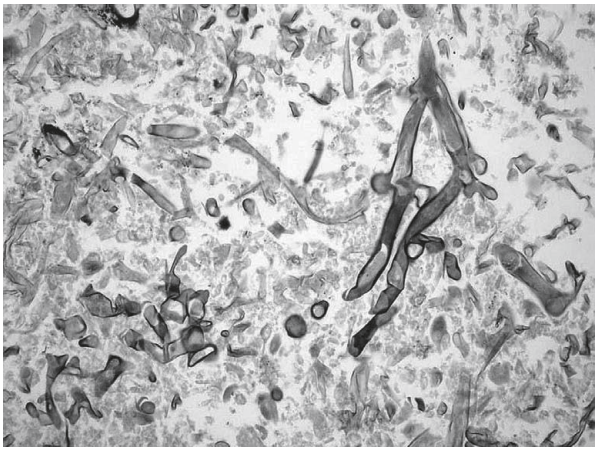

Fig. 4 — Photomicrograph shows numerous broad diameter, nonseptate hyphae with branches occurring at right angles (periodic acid-Schiff; original magnification, 400×).

cavity. Unsurprisingly, hard stool was found in the sigmoid colon which caused partial obstruction. The patient then underwent a right hemicolectomy. She was transferred to the intensive care unit and her condition stabilized.

However, on the third postoperative day, the patient developed acute upper gastrointestinal bleeding with coffee-ground aspirate from the nasogastric tube. Upper endoscopy revealed one mass measuring 4 cm at the angle of the stomach. Additionally, a separate 5-cm mass was seen in the posterior wall of the body. Some A2 ulcers were seen on the surface of the masses (Figs. 1 and 2). Histopathological evaluation demonstrated numerous broad, nonseptate hyphae with right-angle branched fungi with hematoxylin and eosin as well as silver stains in the exudates within the ulcer, which suggested mucormyces (Figs. 3 and 4). She received amphotericin B 0.5 g/kg/day for 28 days, which was discontinued when follow-up endoscopy showed complete resolution of the gastric masses (Fig. 5). The patient's subsequent hospital course was uneventful and there was no evidence of recurrent ulcers or bleeding.

3. Discussion

Zygomycoses are infections caused by fungi of the class Zygomycetes, which is composed of the orders Mucorales and Entomophthorals [3]. The term *mucormycosis* is familiar and used by most clinicians as it is widely used in reports in the medical literature. In addition, it is used as an indexing term within medical databases.

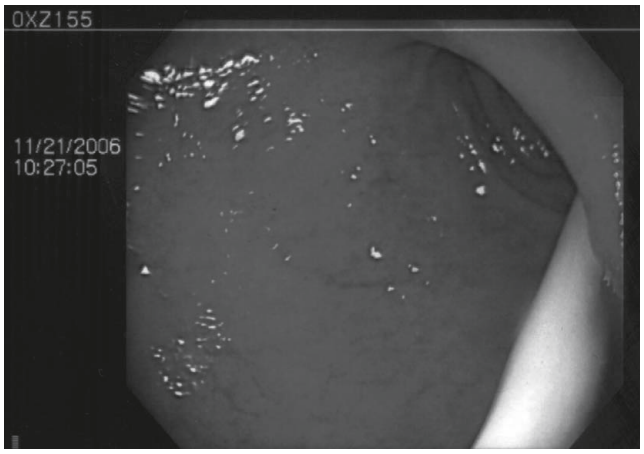

Fig. 5 — Endoscopy reveals complete resolution of gastric masses after amphotericin B treatment.

Mucormycosis usually involves rhinocerebral infections which account for more than half of the cases [1]. Gastrointestinal tract infections are rare, and affect only 7% of all zygomycosis cases [4]. In non-transplant patients, the stomach is the site most frequently involved (67%), followed by the colon (21%), small intestine (4%), and esophagus (2%) [5]. Gastric mucormycosis is primarily found in patients with diabetes mellitus, solid organ transplantation, corticosteroid use and malnutrition. Cases associated with acquired immunodeficiency syndrome and iatrogenic outbreaks and those that followed use of wooden tongue depressors have been reported [6,7].

After reviewing the present case, alcoholism seems to be one of the risk factors of gastric mucormycosis which is seldom described in published reports in the medical literature. We propose the following explanation for the pathogenic basis of alcoholism in gastric mucomycosis. Firstly, it is known that neutrophils are the prominent components of the host immune system that respond to the Mucorales [1]. Secondly, oxidative metabolites that are generated by phagocyte respiratory burst have been shown to be fungicidal to *Rhizopus oryzae* hyphae [8]. Thirdly, patients with acute or chronic alcohol abuse have impaired cellular immunity, which is particularly notable with regards to antigen-specific immune response. It thereby, unavoidably, increases the host's susceptibility to infections [9].

Two cases of gastric mucormycosis in patients with alcoholism have been reported [10,11]. Both cases described patients with abdominal pain similar to our patient. One patient had melena and hematemesis and the other appeared toxic with fever and vomiting. Upper endoscopy revealed ulcerated lesions with raised edges and necrotic base contained thick-dirty necrotic debris and hemorrhage clots. Therefore, we suggest that alcoholism might be one of the risk factors for gastric mucormycosis. Also, we would point out that ingesting fungal spores and germination by patients with alcoholism can be harmful, as ethanol may disrupt activation of macrophages and dendritic cells, which play crucial roles in immunity for eliminating fungal spores.

The initial manifestations of gastrointestinal mucormycosis are abdominal pain and distention associated with nausea and vomiting [1]. Fever and hematochezia may also be found. The diagnosis of gastrointestinal mucormycosis is made in accordance with endoscopic biopsy results of the lesions that show the characteristics of broad hyphae, irregular branched and rare septations. The hyphae of the fungi in this case were differentiated from those of *Aspergillus*, because hyphae of *Aspergillus* are narrower with regular branching and many septations. Routine histological stain with hematoxylin and eosin or with special stains such as Grocott's methenamine–silver and periodic acid-Schiff are adequate to identify fungal hyphae. On the other hand, since *Mucor* species are potentially angioinvasive, infarction and necrosis of host tissues are seen as the hallmarks of invasive disease.

Because of its rapidly progressive and fatal clinical course, the crucial medical intervention for mucormycosis includes correcting underlying diseases, removing necrotic tissue by surgery and using antifungal agents promptly. Meanwhile, amphotericin B deoxycholate and its lipid derivatives remain as the standard therapy for mucormycosis. The recommended dose of amphotericin B deoxycholate is 1 to 1.5 mg/kg/day. As to other newer antifungal agents, such as voriconazole and caspofungin, they are not effective against mucormycosis. Furthermore, a novel extended-spectrum investigational triazole named posaconazole has a potential role in the treatment of zygomycosis that is refractory to the standard therapy. In a clinical trial of 91 patients with either proven (n=69) or probable (n=22) infections with zygomycosis who failed or could not tolerate standard therapy, posaconazole achieved either complete or partial response in 60% of patients, while 21% were stabilized [12].

Previously researchers have reported that the mortality rate of those with gastric mucormycosis approaches 98% [13]. Hence, prompt diagnosis, reversal of predisposing condition, aggressive surgical debridement and early use of amphotericin B are the cornerstones of therapy for this fatal disease. Astute clinicians should also recognize alcoholism as a risk factor for gastric mucormycosis.

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