

A Case of Gastric Zygomycosis in a Diabetic Patient Successfully Treated with Total Gastrectomy

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ABSTRACT

Zygomycosis is regarded as a rare fungal infection in diabetics and other immunocompromised patients. The usual manifestations of this infection are: rhinosinusitis, pansinusitis, rhino-orbital and rhinocerebral. Primary gastrointestinal (GI) zygomycosis is a rare disease with a high mortality rate. The stomach is the most common site involved in GI mucormycosis. There are few reported cases of GI zygomycosis in the literature. Here we report a case of a diabetic woman with abdominal pain secondary to gastric zygomycosis that successfully responded to surgical treatment.

KEYWORDS

Zygomycosis; Diabetes; Gastrectomy

INTRODUCTION

Zygomycosis is a rare opportunistic fungal infection caused by various members of the class Zygomycetes, especially Mucorales, which is subdivided into the genera *Absidia*, *Rhizopus* and *Mucor*.^{1,2} They are typically found in soil, spoiled foods, bread and dust, therefore, most individuals are exposed to these fungi on a daily basis but most often diabetics or immunocompromised patients are susceptible to this infection.³

The common manifestations of zygomycosis are rhinosinusitis, pansinusitis, rhino-orbital and rhinocerebral.

Although uncommon, the gastrointestinal (GI) tract can also be involved, with the stomach most commonly affected with variable signs and symptoms such as

gastric ulcers, gastritis and gastric perforation.^{4,5}

GI zygomycosis is associated with a high mortality rate and early diagnosis is critical. In the past when appropriate diagnostic tools were not available, mucormycosis was frequently found by autopsy.⁶

The advance in current endoscopic technology has increased diagnostic rates and made successful management available with appropriate treatments such as debridement of contaminated tissues. Herein, we report detailed clinical and pathologic findings of a woman with the diagnosis of gastric zygomycosis.

CASE REPORT

A 36 year-old diabetic woman referred with chief complaints of 20

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day epigastric pain, bilious vomiting, weight loss, fever and constipation. The patient was known to have diabetes and hypothyroidism, and used levothyroxine and glibenclamide.

On examination she was thin, febrile, with mild tenderness in the epigastric region of her abdomen. Laboratory tests showed white blood count of 16000 mm³; hemoglobin, 8.8 gr/dL, platelets, 250000; and elevated amylase level of 726 u/L. Plain radiography showed a distended stomach.

Endoscopy was performed which disclosed an extensive sub-mucosal hemorrhage, severe congestion with a snake skin appearance, and a 10×15 cm ulcerated lesion involving the proximal part of the greater curvature of the stomach. Biopsy specimens demonstrated only necrotic material and acute inflammatory cells that contained numerous fungal elements. The fungi were broad, aseptate with acute angle branching (Figure 1).

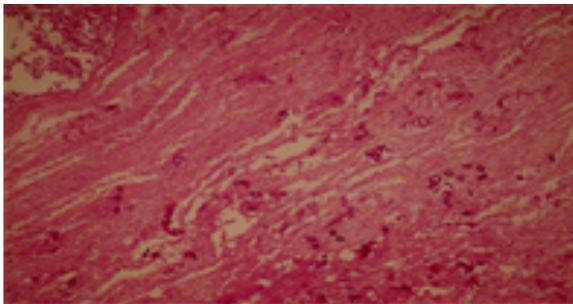


Figure 1: Histopathological finding. Microscopic view of the biopsy specimen shows several fungal hyphae (H&E, X400)

Due to the biopsy result and aggravation increased of abdominal pain, laparotomy was performed which showed gray/brown rubbery necrotic tissue in the stomach that covered a 10×15 cm defect in the wall of gastric's body (Figure 2).



Figure 2: Gross specimen. A gray brown rubbery and necrotic tissue in the stomach covering a 10×15 cm defect in the wall of body of stomach.

No other intra-abdominal pathology was found. A total gastrectomy with Roux-en-Y esophagojejunostomy and surgical debridement of necrotic tissue were performed and the specimens were sent for pathologic evaluation. Histopathology of the stomach showed extensive ulceration and acute inflammatory cells in the entire thickness of the organ.

Numerous characteristic non-septate fungal hyphae were seen within the inflammatory exudates region, which showed a superimposed infection that was located on a pre-existing peptic ulcer. Focal vascular invasion was also present.

Post-operative investigations showed no other sites of infection. The patient was treated with amphotericin B lipid complex (1 mg/kg/dose). She showed significant improvement and at follow up four months after surgery remained clinically well and had gained weight.

DISCUSSION

We present a case of gastric zygomycosis, which is an extremely rare site for this infection whose outcome is usually fatal.^{4,5} Zygomycosis is most often seen in diabetics and immunosuppressed patients and caused by various members of the Phycomycetes, such as *Rhizopus*, *Mucor*, and *Absidia*.^{1,2} This infection has a high mortality rate of 79–100%.^{6,7}

Fortunately our patient was successfully treated with the combined approach of early diagnosis, correction of the underlying predisposing condition, surgical debridement and early systemic amphotericin therapy.

Several studies have shown that antifungal treatments are insufficient to treat this condition.^{8,9} Thus, it is very important to consider surgical treatment followed by medical therapy.

Amphotericin, preferably the liquid formulation, is the choice of antifungal treatment because an adequate dose can be delivered.⁸ There is no renal toxicity, particularly in diabetics. The common prophylactic antifungal agents such as fluconazole and itraconazole are not effective

against this fungus.¹⁰ Newer clinical and *in vitro* investigations have shown that antifungal drugs such as voriconazole and caspofungin also are ineffective against mucor.^{10,11}

Petrikkos et al. have reported their experience on 24 patients with mucormycosis at their centre. They had no GI zygomycoses in their series.

Seven patients did not undergo surgical debridement and all died. They concluded that the degree of immunosuppression and surgical treatment are the two most important factors which contribute to a good outcome.¹²

Zygomycosis is usually misdiagnosed.¹³ This delay in diagnosis often leads to a catastrophic dissemination and inevitable death. Because of multiple sites of involvement in these patients, the surgical option may not be feasible.

However early medical and surgical treatment, as we did in our patient, could prevent further dissemination.

In summary, we report an extremely rare form of zygomycosis that responded to combined surgical and medical treatment; we believe that this was partly a result of the early diagnosis and treatment. It should be kept in mind that medical treatment alone in GI zygomycosis is always unsuccessful. The combined antifungal treatment and surgical removal of the involved area is the key to success.

CONFLICT OF INTEREST

None declared.

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