Case Report

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Mucormycosis presenting as gastric perforation peritonitis in a malnourished young adult: a rare case report

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ABSTRACT

Mucormycosis is a rare infection which is largely diagnosed in immune-compromised patients. The infection can cause pulmonary, rhinocerebral, skin and soft tissue, central nervous system, gastrointestinal and disseminated disease, with gastrointestinal involvement being the rarest presentation. Outcome and mortality of zygomycosis varies with the underlying condition and site of infection, it is however very high in general. Diagnosis is usually delayed and delay in initiation of amphotericin B treatment leads to poor outcome. We report rare case of a malnourished young adult who presented with gastric perforation peritonitis due to mucormycosis infection.

Keywords: Gastric perforation, Gastrointestinal, Mucormycosis

INTRODUCTION

Mucormycosis is a rare, opportunistic fungal infection led by fungal agents within the order *Mucorales*; occurs almost solely in immunocompromised hosts, such as patients with diabetes mellitus, burns, malnutrition, leukemia, lymphoma, septicemia, renal disease, and following long-term treatment with steroids and antibiotics; and is associated with high mortality rates. ¹⁻³

Nevertheless, 19% of patients have no underlying condition at the time of infection.

This rare angioinvasive fungal infection, mucormycosis, can cause infections that are rhinocerebral, pulmonary, cutaneous, gastrointestinal, renal, or disseminated, but primary gastrointestinal infection is an uncommon clinical presentation with a prevalence of only 7% of reported cases.⁴

All portions of the gastrointestinal system can be affected by infection with mucormycosis.⁵ In patients found to

have gastrointestinal invasive mucormycosis, the common site of infection is the stomach, ileum and colon.⁵ Here, we report rare case of a malnourished young adult who presented with gastric perforation peritonitis due to mucormycosis infection.

CASE REPORT

A 15 year old boy presented to the surgical emergency with complaints of abdominal pain and distension for 1 day with history of bilious vomiting and not passing stools/flatus for past 2 days. He was recently diagnosed with pyogenic (bacterial) meningitis 5 weeks before for which he was undergoing treatment as a result of which he was chronically hospitalized and malnourished. He had no other co-existing illnesses or comorbidities. On examination, he was conscious, with altered mental status, poorly nourished, emaciated and had pulse rate of 115 bpm, BP-109/60 mmHg. Per abdominal examination revealed tense, distended abdomen with diffuse guarding and rigidity and absent bowel sounds. His blood reports were unremarkable and he was negative for HIV,

hepatitis B and C viral markers. After initial resuscitation, patient was taken up for an erect abdominal x-ray which revealed free air under right hemi-diaphragm. He was taken up for exploratory laparotomy in view of perforation peritonitis. Intra-operatively, there was around 500 ml bilio-purulent contamination in the peritoneal cavity with a giant gastric perforation of about 2.5×2.5 cm along the greater curvature of the stomach in the anterior aspect shown in Figure 1.



Figure 1: 2.5×2.5 cm perforation on anterior surface of stomach.

Figure 2 shows repair of the perforation was done after edge biopsy of the perforation margin using healthy omentum. Nasogastric and nasojejunal tubes were placed intra operatively under vision. Post operatively the patient was under continuous and intensive vital care.



Figure 2: Omental patch repair of perforation.

The biopsy revealed extensive areas of transmural necrosis with dense acute inflammation and presence of numerous fungal hyphae (PAS positive) morphologically consistent with gastric mucormycosis shown in Figure 3.

He was started on liposomal amphotericin B. None of the other cultures (blood, urine, CSF) revealed mucormycosis

or any other fungi. He had an uneventful post-operative recovery. He was started on nasojejunal feeds on POD4 and was fully orally allowed from POD7. He regained his appetite and his mental status also improved commendably and he was discharged on POD14.

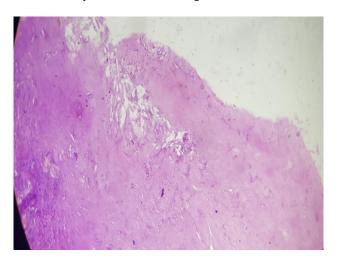


Figure 3: Histopathology showing transmural necrosis with dense acute inflammation and presence of numerous fungal hyphae (PAS positive) morphologically consistent with gastric mucormycosis.

DISCUSSION

The most frequent forms of mucormycosis presentation are sinus (39%), pulmonary (24%), cutaneous (19%), cerebral (9%), gastrointestinal (7%), disseminated (3%), and kidney (2%).¹⁻⁴ Among the several forms, gastrointestinal mucormycosis is rare, and the manifestations vary from the colonization of peptic ulcers to infiltrative disease with vascular invasion and dissemination.²

In gastrointestinal involvement, the most frequently compromised organ is the stomach (58%), followed by the colon (32%), small intestine, and esophagus. The incidence is increasing, and the diagnosis carries a significant mortality rate of up to 85% due to perforation and massive bleeding. 1.2.4

Risk factors for mucormycosis include diabetes mellitus, diabetic ketoacidosis, neutropenia, corticosteroid use, hematologic malignancies, bone marrow or solid organ transplantation, treatment with deferoxamine, iron overload, and human immunodeficiency virus infection and acquired immune deficiency syndrome (HIV/AIDS). Predisposing factors for gastrointestinal mucormycosis have been reported to include kwashiorkor, malnutrition, pellagra, uremia, amebic colitis, and typhoid fever.

The diagnosis of gastric mucormycosis is made on biopsy that shows aseptate, broad PAS-positive fungal hyphae adjacent to the necrotic areas. Definitive diagnosis by microbiological culture and identification of the causative organism to the genus and species carries valuable epidemiological, therapeutic and prognostic implications. Polyene antifungal agent, amphotericin B, is typically active against the fungus, and liposomal formulations have been effective due to less nephrotoxicity. Liposomal amphotericin B can be given at 5-15 mg/kg/day and the successful therapy course usually lasts for a period of

between 4-6 weeks.⁷ Posaconazole is a new triazole antifungal agent that has recently been approved by the US food and drug administration and is typically prescribed at a dose of 400mg, twice daily. Combination therapy can be used, as has been previously reported in 2006 by Rickerts et al as a successful treatment option in patients with disseminated mucormycosis who were unable to undergo surgery.⁸

Table 1: Recent similar case reports involving gastric mucormycosis.

Study group	Age and sex	Comorbidities	Presentation	Findings	Treatment	Outcome
Study patient	15 M	Chronic malnurtrition ICU admission due to meningitis	Acute surgical abdomen	2.5×2.5 cm gastric perforation along the greater curvature	Primary repair of perforation Liposomal amphotericin B	Survived
Sánchez- Velázquez et al ¹	53 F	Prolonged ICU admission Aspiration pneumonia	Massive upper GI bleeding Hypovolemic shock	Perforated gastric ulcer at GE junction and fundus	Total gastrectomy without reconstruction due to coagulopathy	Died
Kulkarni RV et al ³	50 M	Alcoholic and DM	Acute surgical abdomen	4×4 cm perforated ulcer in the gastric body	Wedge resection of the ulcer	Died
Alvarado- Lezama et al ¹⁰	53 M	DM ICU admission due to head trauma	Diabetic ketoacidosis Upper GI bleeding	CT showed emphysematous stomach erosive esophagitis, necrotizing gastritis	Total gastrectomy	Died
Song et al ¹¹	60 M	History of AML malnutrition	Sepsis CT showed discontinuity of posterior wall of the stomach	Laparotomy showed massive gastric bleeding and multiple perforations	Total gastrectomy and splenectomy liposomal amphotericin	Survived

However, antifungal therapy alone is typically inadequate, and surgery to debulk or resect all infected tissue is often required for an effective cure. Roden et al, in his review, has confirmed the importance of multimodality treatment with surgery and amphotericin B.⁹

CONCLUSION

Gastric perforation due to mucormycosis is a rare occurrence. Knowledge of the properties of this fungal pathogen and awareness of its clinical presentation and the patient risk factors for mucormycosis infection may benefit the early diagnosis and management of this disease. A high degree of suspicion is required to make the diagnosis of mucormycosis, and prompt and aggressive surgical debridement with initial adjunctive treatment with liposomal amphotericin B therapy is recommended to improve patient outcome.

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REFERENCES

- 1. Velázquez VP, Pera M, Gimeno J, Zapatero A, Nolla J, Pera M. Mucormycosis: an unusual cause of gastric perforation and severe bleeding in immunocompetent patients. Rev Esp Enferm Dig. 2017:109(3):223-5.
- 2. Jung JH, Choi HJ, Yoo J, Kang SJ, Lee KY. Emphysematous gastritis associated with invasive gastric mucormycosis: a case report. J Korean Med Sci. 2007;22(5):923-7.
- 3. Kulkarni RV, Thakur SS. Invasive gastric mucormycosis-a case report. Indian J Surg. 2015;77(1):87-9.
- 4. Kontoyiannis DP, Lewis RE. Invasive zygomycosis: update on pathogenesis, clinical manifestations, and management. Infect Dis Clin North Am. 2006;20:581-607.
- Prasad SBN, Shenoy A, Nataraj KS. Primary gastrointestinal mucormycosis in an immunocompetent person. J Postgrad Med. 2008;54:211-3.

- 6. Spellberg B. Gastrointestinal mucormycosis: an evolving disease. Gastroenterol Hepatol (NY). 2012;8:140-2.
- 7. Petrikkos G, Skiada A, Sambatakou H, Toskas A, Vaiopoulos G, Giannopoulou M, Katsilambros N. Mucormycosis: ten-year experience at a tertiary-care center in Greece. Eur J Clin Microbiol Infect Dis. 2003;22(12):753-6.
- 8. Rickerts V, Atta J, Herrmann S. Successful treatment of disseminated mucormycosis with a combination of liposomal amphotericin B and posaconazole in a patient with acute myeloid leukemia. Mycoses. 2006;49(1):27-30.
- Roden MM, Zaoutis TE, Buchanan WL, Knudsen TA, Sarkisova TA, Schaufele RL, Sein M, Sein T, Chiou CC, Chu JH, Kontoyiannis DP. Epidemiology and outcome of zygomycosis: a

- review of 929 reported cases. Clin Infect Dis. 2005;41:634-53.
- Alvarado-Lezama J, Espinosa-González O, García-Cano E, Sánchez-Córdova G. Emphysematous gastritis secondary to gastric mucormycosis. Cir Cir. 2015;83(1):56-60.
- 11. Song KY, Kang WK, Park CW, Choi YJ, Rha SE, Park CH. Mucormycosis resulting in gastric perforation in a patient with acute myelogenous leukemia. Surg Today. 2006;36:831-4.

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