Mucormycosis-unusual cause of lower GI bleeding: A rare case report

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**Abstract**

**Introduction:** Mucormycosis is a rare systemic fungal infection seen in immune-compromised patients. Gastrointestinal tract involvement is not usual.

**Case presentation:** A 36 years female presented with fever and progressive bilateral leg swelling for 25 days. She was diagnosed as nephrotic syndrome and started on methylprednisolone and cyclophosphamide. She developed hematochezia during hospital stay. On colonoscopy, ulceroproliferative lesion was noted in caecum.

**Discussion:** Mucormycosis is an opportunistic angioinvasive disease caused by fungus zygomycosis. It is a rare disease and often manifests as a life-threatening condition in immune-compromised patients. Diagnosis is confirmed by histopathological examination and culture. It is usually treated by the anti-fungal drug- liposomal amphotericin and surgical debridement.

**Conclusion:** Mucormycosis is a fatal systemic fungal infection, which can present as lower gastrointestinal bleeding in immunocompromised patients.

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**Keywords:**
- Mucormycosis
- Hematochezia
- Gastrointestinal tract
- Caecum
- Case report

**1. Introduction**

Mucormycosis is a systemic fungal infection caused by the mucor or rhizopus that belongs to the Mucoraceae family. It is a ubiquitous saprophytic mold which grows in soil and organic matter and produces hyphae after inhalation or ingestion. It represents the third most common angioinvasive fungal infection following candidiasis and aspergillosis [1]. It is common in immune-compromised patients like Diabetes Mellitus, Malignancies, HIV/AIDS, Corticosteroid/Immunosuppressant therapy. There are six types of mucormycosis, namely: rhinocerebral, pulmonary, cutaneous, gastrointestinal tract, disseminated, and miscellaneous (endocarditis, osteomyelitis, renal) [2,3]. Stomach is the most common site of involvement in gastrointestinal tract (GI) followed by colon and ileum [4]. GI symptoms occur due to colonization of fungus leading to mucosal ulceration, infiltration and vascular invasion. Clinical progression is rapid and almost universally fatal if untreated [5]. It is diagnosed by histopathological examination or tissue culture [6]. It is treated by systemic antifungal drug and surgical debridement or resection. This case report has been reported in line with the SCARE Criteria [7].

**2. Case presentation**

We report a case of 39 years lady who presented with fever for one and half month, sore throat followed by swelling of bilateral lower limb 25 days back. Swelling gradually progressed to involve face. Then she noticed cola-colored urine with gradual decrease in urine output. She also gave a history of evening rise in temperature associated with chills and rigor. She gave no any past medical, surgical, psychosocial and family history.

Investigation showed Total Leucocyte count of 14,000 with Neutrophils 75%, Lymphocytes 20%, Hb-7 g%, Urine Protein 3+. She was evaluated and treated for Nephrotic syndrome with methylprednisolone in nephrology ward. She developed Acute Kidney Injury (AKI) after 7 days of admission and required hemodialysis (HD). Treatment was escalated to immunosuppressant Cyclophosphamide with Regular hemodialysis (HD) for her Nephrotic syndrome induced AKI.

On 2nd week, there was bleeding per rectum. Proctoscopy followed by Colonoscopy was done for bleeding per rectum which revealed ulceroproliferative growth with friable mucosa in ascending colon and Caecum as shown in Fig. 1, and biopsy was taken in suspicion of malignant pathology.
operative ward for 1 day. Her post operative condition was static with modialysis and amphotericin B after immediate stabilization in post (Fig. 3). Intra operative course was uneventful.

boring next lesion measuring 1.5 cm width was noted in ileocecal junction with neighboR Standard Hemicolectomy with Double barrel ileo colostomy was laparotomy. Intra operatively firm mass was found at ileocecal junction. Contrast enhanced computed tomography (CECT) abdomen and pelvis was normal. We proceeded for exploratory laparotomy. Intra operatively firm mass was found at ileocecal junction. Right Standard Hemicolectomy with Double barrel ileo colostomy was done.

On cut section, circumferential black color ulcerative lesion measuring 1.5 cm width was noted in ileocecal junction with neighboring next lesion measuring 1 x 5 cm size ulcerative lesion in the posterior wall of ascending colon 5 cm distal to IC junction as shown in (Fig. 3). Intra operative course was uneventful.

Post operatively she was managed in Medical ICU with regular Hemodialysis and amphotericin B after immediate stabilization in post operative ward for 1 day. Her post operative condition was static with Glasgow Coma Scale of 15 and daily urine output of less than 100 ml over 24 h every day. On POD 10, there was sudden rise in serum creatinine level following which patient required noradrenaline support to maintain her Blood pressure. Gradually her blood pressure was below normal level despite of vasopressor support. She was reintubated on POD12 There was persistent increase of serum creatinine with metabolic acidosis with blood PH of 7.01 and she succumbed on 14th POD.

3. Clinical discussion

Mucormycosis is an opportunistic angioinvasive fungal infection belongs to zygomycetes family. Zygomycetes are divided into two orders: the Entomophthorales and the Mucorales. Entomophthorales contain rare pathogenic species, whereas Mucorales contain the most common human pathogens specifically Rhizopus, Mucor, Absidia, and Cunninghamhamellaceae. These organisms with low virulence causes disease less frequently and usually present in immunocompromised individuals. The Rhizopus species is the most pathogenic and significantly more virulent than other fungi [5]. Mucormycosis is a rare disease and often life-threatening condition that is commonly seen in patients with risk factors like diabetes, metabolic acidosis, hematological or solid malignancies, neutropenia, trauma, use of corticosteroids, solid organ, or stem cell transplant, iron overload or deferoxamine use, malnourishment, and exposure to newer broad-spectrum antifungals like Echinocandin and Voriconazole like our case was under immunosuppressant [8,9]. Infection can be of rhinocerebral, pulmonary, cutaneous, gastrointestinal, renal, or disseminated type; gastrointestinal infection comprises only 7% of reported cases so can say our case is rare [2]. Invasion of blood vessels by hyphae leads to arterial thrombosis, tissue infarction, hemorrhage and necrosis. Mechanism of GI tract entry is not clear. Fungi pervasive in environment enters in milieus like nasogastric intubation, ingestion of contaminated food.

Clinical features of GI mucormycosis are nonspecific, which includes abdominal pain and distention, nausea and vomiting, hematemesis, hematochezia, and intestinal perforation with peritonitis where our case presented with hematochezia [10]. It may present with bowel obstruction. Biopsy and histopathological examination is the best method to detect mucormycosis, like we found hyphae in HPE of our case. The histopathological hallmark of mucormycosis infection is vascular invasion, causing thrombosis and infarction, or secondary hemorrhages of neighboring tissues; focal areas of granulomatous inflammation are occasionally present [11]. The diagnosis is established histopathologically by the presence of characteristic broad, branching and non-septate hyphae in infected tissues, usually in connotation with angioinvasion, vascular thrombosis and infarction like in our case there was similar features of fungi without identifiable vascular invasion. Histologically it has been categorized into colonization, infiltrative and invasive as in our case its colonized type [6].

Recently published joint clinical guidelines from the European Society for Clinical Microbiology and Infectious Diseases and the European Confederation of Medical Mycology strongly recommend Liposomal amphotericin B as drug of choice and recommend dose is 5 mg/kg daily; the use of amphotericin B deoxycholate was discouraged because of its nephrotoxic side effects [12].

Besides to antifungal therapy, early surgical debridement improves survival (62% with antifungal alone, 57% with surgery alone, and 70% with both) [5].

However, our case could not be recovered despite resection of diseased part and use of systemic antifungal, amphotericin B.

4. Conclusion

Gastrointestinal mucormycosis is a fatal disease if not treated in time and aggressively. GI tract Mucormycosis could cause intra luminal bleeding. It could differential diagnosis for GI tract bleeding in patients with high-risk factors though it is rare. Early diagnosis, prompt administration of antifungal therapy and surgical resection of infected and necrotic tissue decrease morbidity and mortality of disease burden.
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Fig. 3. Cut section of the specimen showing ulcerative lesion around ileo-cecal (IC) junction.

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Gyaneswhor Shrestha: Study concept, data collection surgical therapy for patient.
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Declaration of competing interest
None.

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