Isolated gastrointestinal mucormycosis mimicking peptic ulcer disease
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Citation

Abstract
Mucormycosis is a relatively uncommon opportunistic fungal infection caused by Zygomycetes; diagnosis of which is usually made during histopathological examination. We present an autopsy case of a non-diabetic patient who clinically was diagnosed to have a perforated gastric peptic ulcer. On histopathological examination the whole of gastrointestinal tract showed mucor hyphae in the wall leading to extensive infarction and gastric perforation. Other organs showed no evidence of infiltration by the fungus. Diagnosis of isolated gastrointestinal tract mucormycosis was made, which is very rare. Greater awareness of the clinical settings in which the gastrointestinal mucormycosis develops and its early detection with appropriate treatment can reduce the high fatality associated with this infection.

INTRODUCTION
Mucormycosis (synonymous with phycomycosis and zygomycosis) is caused by fungi of the class Zygomycetes, order Mucorales and mainly by species of genera Rhizopus, Mucor and Absidia. It is an opportunistic fungal infection seen in uncontrolled diabetes and other chronic debilitating diseases. The incidence of the disease is gradually increasing as a result of wide spread use of antibiotics, steroids and immunosuppressive therapy. Diagnosis is usually made during histopathological examination of the autopsy material by the presence of broad, non-septate mycelia in tissues. We here present an autopsy case of a non-diabetic patient with isolated gastrointestinal mucormycosis who was clinically thought to have perforated gastric peptic ulcer.

CLINICAL HISTORY
A 50 year-old male-patient, non-diabetic, non-hypertensive, with no drug history came with complaints of abdominal pain of one and half months duration. The pain was colicky and diffuse in nature, with no aggravating or relieving factors. There was associated gradual distension of abdomen and vomiting on and off. On admission the general condition of the patient was poor and was anuric. Systolic blood pressure was 70 mm of Hg. Laboratory examination revealed mild anaemia, a total white blood cell count of 4100/ mm³ and peripheral smear showed mild lymphocytosis. Amongst renal function tests, blood urea nitrogen was 34 mg/dl and serum creatinine of 3.4 mg/dl.

Serology for human immunodeficiency virus was not performed and he had no nasal complaints. He was taken for emergency exploratory laparotomy with clinical suspicion of gastric peptic ulcer perforation. A 1-cm gastric perforation in the anterior wall of pylorus with pus flakes on the serosa of the rest of the intestines was present. The perforation was sutured with the omental patch. The patient died 32-hours post-op.

A complete autopsy was performed on a poorly built and malnourished individual. External examination including the head, face and neck examination was unremarkable. In-situ examination of abdominal cavity revealed dilated and dusky bowel loops with pus flakes on the serosal aspect of the intestines, liver, spleen and diaphragm. (Figure 1)
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The operative site was identified which was intact. Histopathology revealed extensive ulceration of the gastrointestinal tract mucosa, infarction and transmural infiltration by broad non-septate, non-branching fungal hyphae with moderate inflammation. At the perforation site in the stomach, there was dense transmural neutrophilic infiltrate and mucor hyphae were seen reaching upto serosa. Corresponding to the pus flakes on the serosa of the intestines, microscopically there were hyphae and surrounding neutrophils. (Figure 2)

DISCUSSION

Zygomycetes are ubiquitous fungi, often present in soil and decaying organic material. They cause infection only in immuno-compromised states and mucormycosis is commonly associated with diabetic ketoacidosis, immunosuppression following organ transplantation, severe malnutrition, haematological malignancies, chronic debilitating diseases and desferrioxamine therapy in patients requiring haemodialysis. They invade the deep tissues via inhalation of airborne borne spores, percutaneous inoculation or ingestion. Five forms of the disease have been recognised as rhinocerebral, pulmonary, cutaneous, gastrointestinal and disseminated. Rhinocerebral form is common in diabetics and pulmonary and disseminated forms are seen with haematological malignancies, whereas gastrointestinal infection is thought to arise from the ingestion of spores and has been traditionally linked to...
extreme malnutrition. This could be the case with our patient who was poorly built and malnourished.

Mucormycosis confined only to gastrointestinal is uncommon and accounts for only 7% of the cases. When the gastrointestinal tract is involved, the stomach is the most common site and rarer intestinal forms have a predilection for the terminal ileum and caecum. The stomach is involved in 67% of all gastrointestinal manifestations with a reported mortality rate of 98%. It is followed by colon (21%), small intestine (4%) and oesophagus (2%). In a series of seven cases of invasive gastric mycosis reported in literature, all presented with perforated viscus either intraoperatively or at autopsy. In a study of gastrointestinal mucormycosis of 58 cases only one case showed involvement of stomach, small and large intestine. In another study of 14 cases of only intestinal mucormycosis after excluding gastric forms, gastric involvement was seen in only one case along the involvement of small and large intestines. In our case, the entire length of the gastrointestinal tract was involved which makes this unusual even amongst the gastrointestinal form. Though some of the reported cases have been in diabetics, in the literature there is no clear association of diabetes with isolated gastrointestinal mucormycosis as is with rhinocerebral forms.

The disease is reported both in adults and in neonates and infants where it can present as necrotising enterocolitis and intestinal perforation. In a study of cases of gastrointestinal mucormycosis, 5 were premature babies and prematurity was also postulated as risk factor for this infection. The usual symptoms of gastrointestinal mucormycosis include vomiting, bloody diarrhoea, upper gastrointestinal bleeding, abdominal lump and abdominal distension. This patient presented with pain in abdomen and gradual distension of abdomen. In a case report describing the upper gastrointestinal endoscopy findings, the lesions appeared as ulcerated plaques with raised edges and were covered with black necrotic slough. Limited reports in literature are available describing the radiologic findings of mucormycosis. Contrast enhanced CT studies may show diffuse circumferential wall thickening with areas of both intense and poor contrast enhancement in intestinal wall correlating with areas of congestive changes and of necrosis and infarction caused by fungal proliferation respectively. The clinical diagnosis is difficult as the symptoms are non-specific and antemortem diagnosis can be achieved by demonstrating broad, non-septate hyphae with right angle branching in histopathology in the resection or biopsy material.

Mucormycosis can be just colonisation of peptic ulcers in stomach to infiltrative disease or vascular invasion with dissemination. Fungal elements at the base of chronic peptic ulcers may enhance the degree of necrosis and that these cases have protracted disease and deeper ulcers with more perforations. Common to all forms of mucormycosis is vascular invasion with production of necrotic tissue. But in our case we do not think that it is a peptic ulcer complicated by mucormycosis as rest of the gastrointestinal tract was also showing gangrenous changes due to invasive mucormycosis.

Successful treatment consists of early diagnosis, intensive systemic antifungal therapy with amphotericin B and surgical resection. Only non resectional surgery carries high mortality whereas surgery followed by antifungal therapy provides improved survival. In this case, the clinicians’ diagnosis was peptic ulcer disease and hence the line of management of only non resectional surgery did not help the patient. Hence a high index of suspicion is required along with histopathological confirmation for diagnosis of isolated gastrointestinal mucormycosis and greater awareness of the clinical settings in which it develops it a pre-requisite to improved survival and reducing the high mortality in such cases.

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