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Case Report

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# INVASIVE GASTROINTESTINAL MUCORMYCOSIS: A RARE PRESENTATION

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## ABSTRACT

Gastrointestinal mucormycosis is uncommon manifestation with non-specific presentation so diagnosis is delayed which lead to higher mortality. Only the identification of the fungi on histopathological sections caused the diagnosis to be made and appropriate therapy to be initiated at early stage. It highlights the need to keep a high index of suspicion for mucormycosis. Here we diagnosed a case of gastrointestinal mucormycosis with non-specific symptoms and absence of known risk factors in 50 years old male.

**KEYWORDS:** Mucormycosis; Gastrointestinal; mesentery nodules.

## INTRODUCTION

Zygomycoses are parasitic diseases caused by Mucorales species, are those of the genera Rhizopus Lichtheimia, and Mucor. The three most common primary clinical manifestations of mucormycosis are rhinocerebral, pulmonary, and cutaneous infections.[1] Various risk factors have been recognized with mucormycosis. Vascular invasion with tissue infarction is responsible for higher mortality. Gastrointestinal mucormycosis has accounted for 4%-7% of all cases. If diagnosis is delayed the mortality rate is higher and most of cases diagnosed on postmortem examination.[2]

## CASE REPORT

A fifty years old male came to emergency department with complaint of non-specific pain abdomen which was initially present in umbilical region and then progressed to involve entire abdomen. At that time a provisional diagnosis of renal colic was suspected. Routine investigations along with USG abdomen were advised along with supportive treatment. Complete blood count revealed leucocytosis with mild neutrophilia. Urine examination was within normal limits. USG abdomen revealed normal study of organs. Patient was put on antispasmodics, anti-inflammatory and antacids and was discharged.

Patient reported back to emergency department within 2 days with aggravated pain in abdomen and had 2 episodes of vomiting which was greenish in color. Repeated USG Abdomen revealed telescoping of one gut loop in another, along with multiple surrounding lymph nodes in left iliac fossa. (? Intussusception). Contrast enhanced computed tomography (CECT) abdomen was advised. CECT abdomen showed a heterogeneously enhancing soft tissue lesion seen in right side of the pelvis, measuring 45 x 39 mm in size. Another soft tissue lesion of size 26 x 28 mm was seen in mesentery in left paraumbilical region. Minimal ascites was seen. Other Abdominal organ and vascular structure appeared unremarkable. Surgical management was advised for diagnostic and therapeutic purpose.

Intra-operatively, two firm masses were identified in mesentery, one was (3x2 cms) adherent to ileum at about 120 cm distal of DJ junction and second was adherent to mesentery 100 cm proximal to IC junction. Thickening of gut was seen at both of adherent part. Due to suspicion of malignancy, segment of small intestine was excised with mesenteric nodules. We received a gut segment measuring 45 cms in length. External surface was covered with exudate. On cutting open, mucosa was unremarkable. Two nodular mass were identified in attached mesentery measuring 5x3.5x2 cms (3.5 cms away from one of the cut end) and 3.5x3x2 cms (6 cms from other cut end). (Fig. 1).

Microscopic examination of intestinal segment revealed unremarkable mucosa. Mixed chronic inflammatory cells comprising of predominantly of eosinophils, lymphocytes and histiocytes infiltrating into serosa and muscular layers with areas of necrosis were identified in intestinal wall. Necrotic areas along with multinucleated foreign body giant cells were also noted in these mesenteric nodules. (Fig 2 & 3) On higher magnification broad non-septate hyphae with right angle branching were observed which were PAS and GMS stains positive, confirming the morphology of mucor. (Fig. 4-6) Vascular invasion by fungal hyphe were also seen in few section. (Fig. 5) With history, none of the predisposing factors for mucormycosis were identified in this case. A Mucormycotic abscess of mesentery was diagnosed. Antifungal drug (amphotericin B) was given and follow up was uneventful.

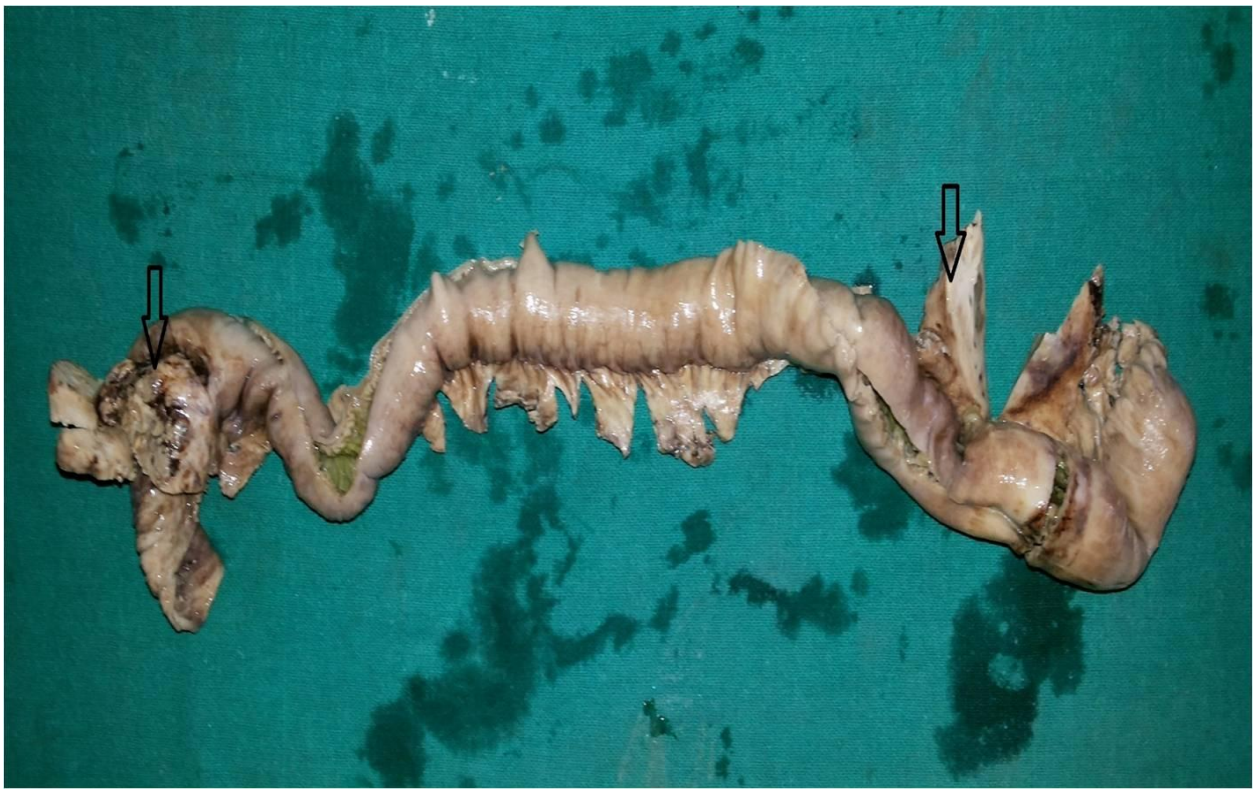

**Fig. 1: Grossly a gut segment measuring 45 cms in length with two nodular mass in attached mesentery.**

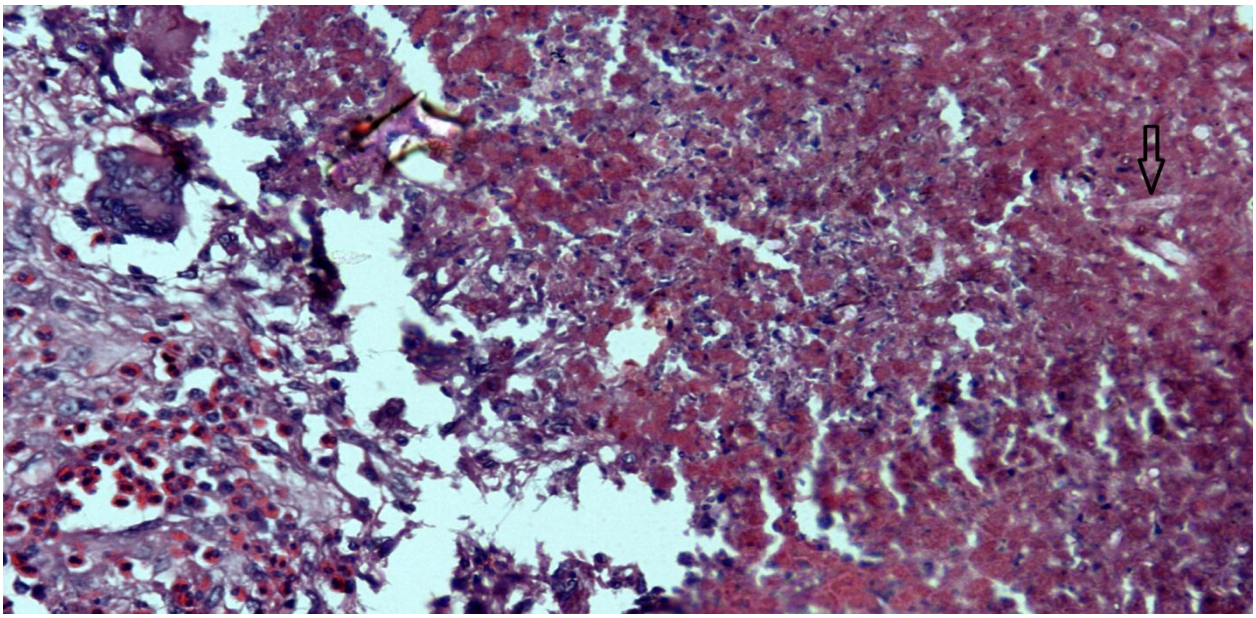

**Fig. 2: Microsection showing collection of eosinophils and giant cells surrounding necrotic tissue and fungal hyphe (arrow). (H&E, 20X).**

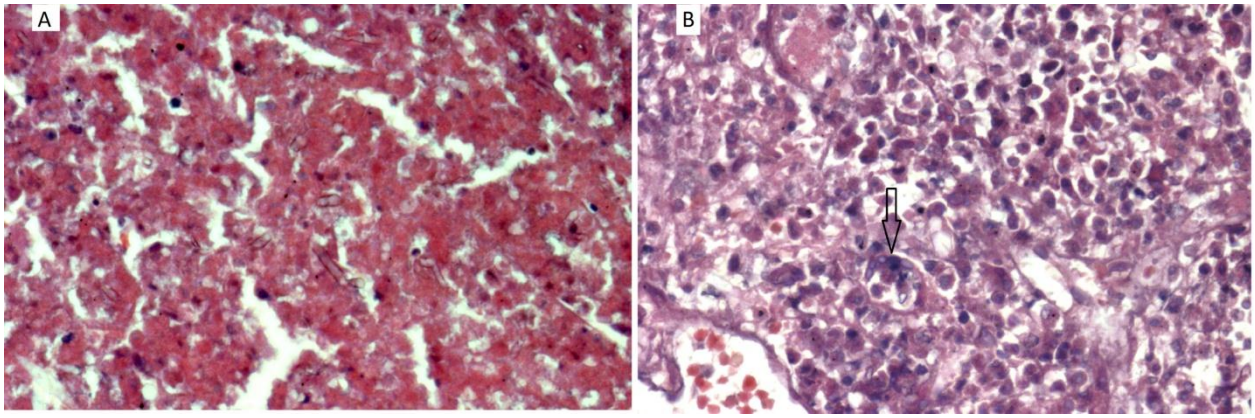


**Fig. 3: (A). Section revealed necrotic debris with multiple fungal hyphe. (H&E, 20X). (B) Microcection showing vascular invasion by fungus. (H&E, 20X).**

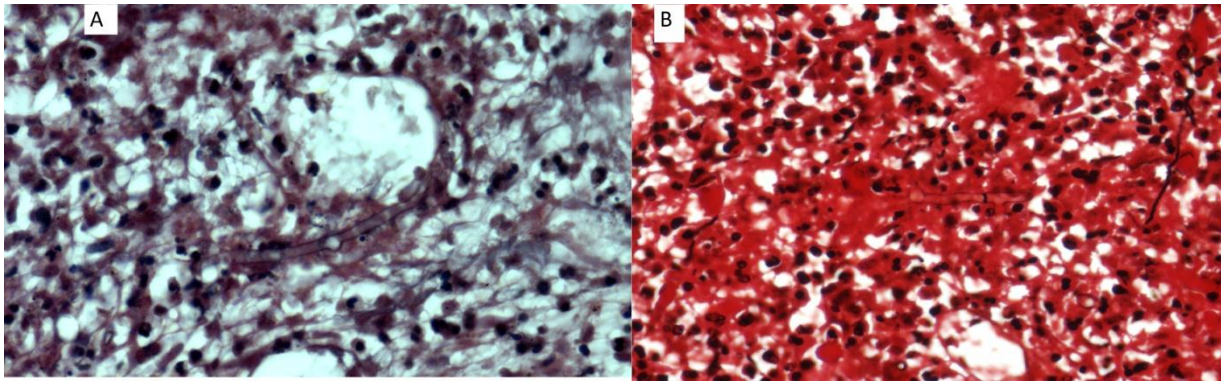


**Fig. 4: (A). PAS stained section confirmed morphology of mucor which is around the blood vessel. (20X) (B) GMS stained section showing acute branching in fungal hyphe. (20X).**

## DISCUSSION

Zygomycoses are uncommon fungal diseases caused by class Zygomycetes. The most of human cases are caused by Mucorales fungi; therefore, the terms mucormycosis and zygomycosis are used interchangeably. The Mucorales species most often recovered from clinical specimens are those of the genera Rhizopus (the most common genus associated with mucormycosis), Lichtheimia, and Mucor. Mucorales are ubiquitous in nature and can be found in soil and decaying matter.[1,2]

Various risk factors have been assoicated with mucormycosis like malignant hematological disease with or without stem cell transplantation, prolonged and severe neutropenia, poorly controlled diabetes mellitus, iron overload, prolonged use of steroids, intravenous drug use, neonatal prematurity and prolonged use of antifungal (voriconazole).[3]

Three routes of infections are inhalation, ingestion and traumatic inoculation. Sporangiospores are the infective forms while angioinvasive hyphal forms are responsible for tissue invasion, infarction and dissemination. The ability to scavenge of free iron from the host is essential for growth and pathogenesis.[1,4] Based on its clinical presentation and anatomic site involvement, invasive mucormycosis is classified into: (1) Rhinocerebral, (2) Pulmonary, (3) Cutaneous, (4) Gastrointestinal, (5) Disseminated, and (6) Uncommon Rare forms, such as endocarditis, peritonitis, and renal infection. Rhinocerebralmucormycosis (ROCM) is the most common form.[4,5]

Gastrointestinal mucormycosis is uncommon presentation which is acquired by ingestion of pathogens in foods such as: fermented milk and dried bread products, fermented porridges and alcoholic drinks and use of spore- contaminated herbal and homeopathic remedies. The diagnosis of gastrointestinal mucormycosis is often delayed because of the non-specific presentation like abdominal pain, distention and vomiting. Sometime infection may present with an abdominal mass (appendiceal, cecal or ileal) mimic as intra-abdominal abscess.[1,2,6]

The stomach is most commonly affected part, followed by the colon and ileum. However, liver, spleen, and pancreas may also involve. The infection usually presents with mass. Due to invasion of blood vessel, bowel perforation, peritonitis, sepsis, and massive gastrointestinal hemorrhage can occur, which are the most common cause of death. Diagnosis of gastrointestinal mucormycosis requires a high degree of suspicion, leading to early use of endoscopic biopsy analysis. If diagnosis is delayed the mortality rate is as high as 85%.[1,2]

The successful management of gastrointestinal mucormycosis requires- early diagnosis and reversal of predisposing risk factors (where possible) along with surgical debridement and initiation of antifungal therapy. There are no recommendations of antifungal agents specific to gastrointestinal infection. Initiation of polyene therapy within 5 days of has been associated with improved survival.[7]

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