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Intestinal Obstruction - Uncommon Manifestation of Gastrointestinal Mucormycosis in a Paediatric Patient: A Case Report

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Abstract: ***Mucormycosis is a dreadful fungal infection commonly observed among immunocompromised individuals but has recently expanded to involve even the immunocompetent patients. It can affect any organ system, seldomly the gastrointestinal tract with incidence of 5-15%. Intestinal obstruction in case of gastrointestinal mucormycosis is an unusual manifestation of an infrequent involvement. A 5-year-old boy presented with sudden altered consciousness, abdominal distension in shock, eventually diagnosed with acute intestinal obstruction and encephalopathy. Subsequent examination of the resected bowel via intestinal biopsy revealed the presence of fungal hyphae. Unfortunately, the patient passed away during the postoperative period. This report is an unusual presentation of gastrointestinal mucormycosis. Despite of its rare occurrence, the devastating nature of the disease yet simple treatment of mucormycosis demands its inclusion among differential diagnosis in such cases. A high clinical scepticism and increased awareness about various presentations of gastrointestinal mucormycosis can benefit in early recognition and prompt treatment.***

Keywords: Mucormycosis, Intestinal obstruction, case report, Ileo-ileal anastomosis, Fungal hyphae

1. Introduction

Gastrointestinal fungal infection, stemming particularly from the zygomycetes class, stands as a non-neoplastic contributor to intestinal obstruction. Within this class, two orders, mucorales and entomophthorales, exhibit varying pathogenic potentials. These commonly inhabit soil and decomposing plant matter, occasionally acting as commensals in the gastrointestinal tracts of certain reptiles, fishes, and amphibians [1]. Mucormycosis can be further classified into pulmonary, rhinocerebral, cutaneous, gastrointestinal, central nervous system, and disseminated type on the basis of organ system implicated. Gastrointestinal mucormycosis is an atypical manifestation which accounts for only 5- 15% cases [2]. However, on most occasions, the diagnosis is made intraoperative or postmortem, hence the precise assessment of incidence is a hurdle [2]. Symptoms may manifest diversely, ranging from vague abdominal pain, vomiting, fever, diarrhoea, abdominal distention and infrequently gastrointestinal bleeding, obstruction, perforation, sepsis and hepatic mucormycosis [2,3]. Recently the disseminated type, a potentially fatal manifestation, has been documented among both immunocompetent and immunocompromised patients, albeit exceedingly rarely. Given its paucity, timely diagnosis may pose challenge and therefore lead to delays, contributing to a notably high mortality rate, particularly among pediatric patients [4,5]. Herein, we present a case of small bowel obstruction resulting from fungal infection in an otherwise healthy child, managed through a combination of surgical and medical approaches.

2. Case report

This report is following the Surgical Case Report (SCARE) guidelines [6]. A 5-year-old Indian boy 2nd in birth order presented with complaints of cough and fever seven days back followed by abdominal distention, pain and vomiting since 4 days, altered consciousness for 3 days. On initial assessment, the child was in shock, with a feeble pulse at rate of 150/min, non-recordable blood pressure, acidotic breathing with respiratory rate of 60 cycles per minute and oxygen saturation of 75% on room air. The patient was resuscitated, intubated and shifted to pediatric intensive care unit (PICU). Ryle's tube insertion and urinary catheterization was ensured. Physical examination revealed a distended and tense abdomen with no evidence of guarding or rigidity. On auscultation bowel sounds were absent.

Laboratory findings indicated a hemoglobin level of 11.5 gm/dl, white blood cell count of 34,200/mm^3 with neutrophilia (89%) and lymphopenia (9%), elevated prothrombin time and INR (19.9 sec and 1.7 respectively), elevated markers of inflammation- ESR-50mm at the end of 1 hour, CRP-55.50 mg/L, D-dimer- 3423.86 ng/ml, ferritin-301ng/ml LDH- 783 U/L. Serological tests for HIV, hepatitis B, and C were negative. Radiological investigations were performed. X ray Chest and abdomen showed multiple air-fluid levels with no evidence of gas under diaphragm (Fig 1).

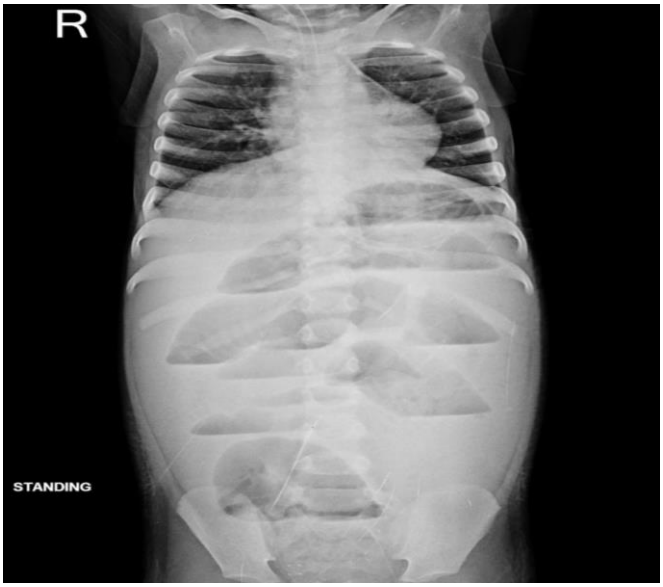

Figure 1: X- Ray chest and abdomen showing multiple air-fluid level.

Ultrasonography of abdomen revealed prominent small bowel measuring 27-29 mm in external calibre filled with fluid and organic matter with sluggish peristalsis.

Following cardiopulmonary stabilization and under coverage of broad-spectrum antibiotics, patient was taken up for emergency exploratory laparotomy. Intraoperatively dilated bowel loops with two impending perforations and sloughed bowel wall segments at respectively 10 and 15 cm proximal to the ileo-caecal junction was observed (Fig 2).

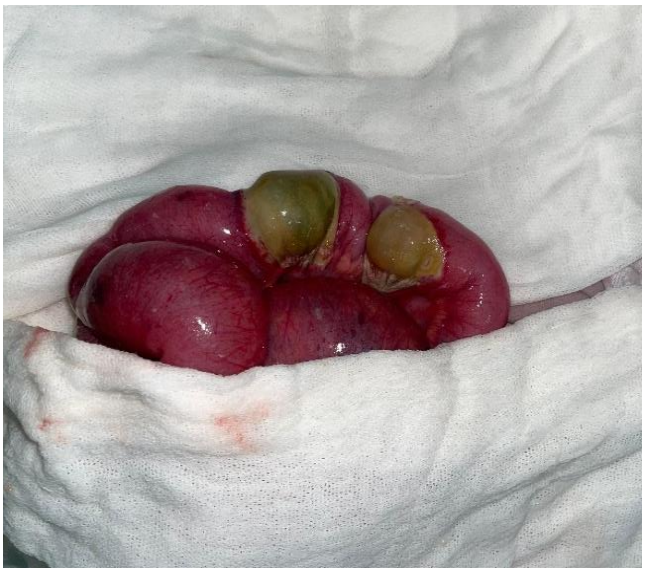

Figure 2: Surgical image revealing dilated bowel loops with two imminent perforations and segments of sloughed bowel wall in the ileum.

Resection of the infected bowel was carried out, followed by ileo-ileal anastomosis. The resected bowel specimen along with lymph nodes was sent for histopathological examination. Postoperatively, child was shifted back to PICU and kept nil per oral with intensive input-output, abdominal girth monitoring on intravenous fluids and RT aspiration every 2 hourly undercover of broad-spectrum antibiotics.

Histopathological examination revealed thinned intestinal mucosal lining, dense inflammatory infiltrate composed of lymphocytes, plasma cells, and neutrophils, with spread into the serosa and pericolic fatty tissue. Lymph node under microscopy unveiled necrosis and inflammatory cell infiltration, along with areas of necrosis and proliferation of congested and dilated blood vessels (Fig 3).

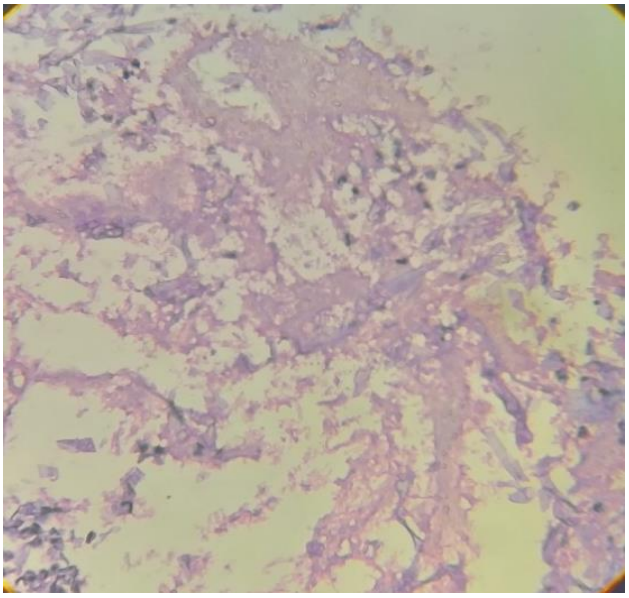

Figure 3: Histopathological examination displaying infiltration of inflammatory cells, areas of necrosis, and proliferation of congested and dilated blood vessels. Along with numerous broad, aseptate fungal hyphae are evident.

Numerous broad, aseptate fungal hyphae were observed, with positive Periodic Acid Schiff (PAS) staining for Mucormycosis. Itraconazole treatment was initiated, but the patient's condition did not improve. Subsequently, he developed acute severe septic shock and pulmonary insufficiency, leading to his demise.

3. Discussion

Gastrointestinal zygomycosis is comparatively infrequent and often fatal, with mortality rates of 40-50%. It can affect any part of the gastrointestinal tract, more commonly the stomach (57.5%), colon (32.3%) and small bowel (7%) mainly ileum [2,7]. They can also infect formerly damaged tissues, and hence widespread disease can develop from trivial gastrointestinal infections. Involvement is usually in form of large ulcers with rolled out irregular edges, often imitating a neoplastic etiology [8,9,10]. Mucorales typically infect immunocompromised patients and have angioinvasive nature leading to tissue lysis due to thrombosis, infarction and enzymatic degradation by fungal proteases, lipases, and mycotoxins. Delayed diagnosis encourages dissemination of disease [1,11]. Conversely, entomophthorales infections, particularly Basidiobolus ranarum, have a predilection for subtropical and tropical regions among mainly immunocompetent individuals. [12,13] Transmission usually occurs via spore inhalation, ingestion of contaminated food, or inoculation after minor trauma, resulting in subcutaneous mycosis, gastrointestinal disease, or systemic lesions [1]. In the case mentioned above, no identifiable transmission risk factors were found. Symptoms of fungal gastrointestinal

disease include abdominal pain, distension, diarrhoea, constipation, fever, hematemesis, hematochezia, weight loss, abdominal distention, hepatomegaly and palpable abdominal masses. Abdominal masses are most commonly found in the colon and rectum, followed by the small bowel, liver, gallbladder and stomach [2,3]. Diagnosis is often retrospective, typically following tissue biopsy during endoscopy or surgical resection. Most cases are initially suspected to be malignancies, inflammatory bowel disease, or diverticulitis [9,10]. Blood tests may reveal peripheral eosinophilia, although it was absent in our case. Fungal culture is the gold standard for diagnosis [9]. Histopathological examinations typically reveal broad, thin-walled, hyposeptate hyphae surrounded by dense eosinophilic material (Splendore-Hoeppli phenomenon) [10]. Recent reports have highlighted the utility of polymerase chain reaction (PCR) and enzyme-linked immunosorbent assay (ELISA) in diagnosing gastrointestinal fungal infections [11]. PCR has demonstrated high sensitivity and specificity; however, its widespread application remains limited due to the rarity of the disease.

The most frequently used antifungal for gastrointestinal fungal infections is itraconazole, followed by amphotericin, ketoconazole, and voriconazole. Voriconazole is preferred for its safety and tolerability, while amphotericin has been associated with clinical failures. Surgical resection combined with antifungal therapy is standard treatment, although successful outcomes with antifungal treatment alone have been reported [12].

4. Conclusion

Gastrointestinal fungal infections can present with diverse range of manifestations including intestinal obstruction, often requiring early surgical intervention. Early initiation of antifungal therapy after confirmatory histopathological examination (HPE) typically leads to favourable outcomes. The diagnostic challenge emphasizes on the importance of raising awareness and promptly recognizing this condition. Without a heightened clinical suspicion and familiarity among clinicians, gastrointestinal mucormycosis may continue to be overlooked, potentially leading to serious consequences.

Consent
Written informed consent to publish his clinical details and images was obtained from the patient.

Conflicts of interest
The authors declare that they have no conflicts of interest.

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