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"An unusual presentation of colonic mucormycosis mimicking carcinoma colon- a surgeon's perspective"

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ABSTRACT

INTRODUCTION: Mucormycosis caused by order mucorales, an ubiquitous saprophytic mold found in soil and organic matter worldwide, is a rare but invasive opportunistic fungal infection. Gastrointestinal mucormycosis is the most uncommon clinical presentation being particularly rare, accounted for 4–7% of all cases.
PRESENTATION OF CASE: We report an unusual presentation of mucormycosis of ascending colon that was simulating carcinoma colon.
DISCUSSION: GI mucormycosis most commonly involves the stomach (57.5%), followed by the colon (32.3%) and the ileum (6.9%). Initial presentations may be abdominal pain and distension, fever, and diarrhoea. Colonic mucormycosis presenting as a mass with altered bowel habit, melena and abdominal pain in our case is extremely difficult to differentiate it from carcinoma colon. A definitive diagnosis of mucormycosis is almost always ascertained by histopathological evidence of fungal invasion of tissue.
CONCLUSION: Knowing these unusual presentations of this disease, surgeon need to maintain a high index of suspicion and perform timely and appropriate diagnostic evaluation to improve patient outcome. Prompt diagnosis, reversal of predisposing conditions, and aggressive surgical debridement remain cornerstones of therapy for this deadly disease.

1. Introduction

Mucormycosis caused by order mucorales [1,2], an ubiquitous saprophytic mold found in soil and organic matter worldwide, is a rare but invasive opportunistic fungal infection. The disease in humans is mainly limited to people with risk factors such as neutropenia [3–6], immune deficiencies [2–4,7,8], malignant disease [2–8], malnutrition [3,4], diabetes [2–8], trauma [2,6], organ transplantation [2–5,7], and iron overload [3–6]. The clinical infection due to mucorales includes rhinocerebral, pulmonary, cutaneous, gastrointestinal and disseminated diseases [3,5].

Gastrointestinal mucormycosis is the most uncommon clinical presentation being particularly rare, accounted for 4–7% of all cases [9]. We report an unusual presentation of mucormycosis of ascending colon that was simulating carcinoma colon.

2. Presentation of case

This study presents the case of a 42 year-old Hindu male, known diabetic with an one-year history of waxing and waning right-sided abdominal pain associated with a change in bowel habits & melena admitted to our hospital with chief complaints of abdominal pain and no passage of stool since two days which was not associated with vomiting or fever. On examination, there was a tender irregular lump of size about 8 × 10 cm felt on right lumbar region extending upto right hypochondrium. Per rectal examination is normal except finger stall being smeared with blood. Other system examination revealed no abnormalities. Laboratory investigations were normal except FBS 186 mg/dl, 1 h PPBS 248 mg/dl and HbA1c 8.5. Ultrasonography of abdomen & pelvis showed thickened, irregular wall of the ascending colon near the hepatic flexure. Upper GI endoscopy revealed normal. On contrast enhanced CT (CECT) of whole abdomen (triple phase), there was an irregular bowel wall thickening (15–19 mm) with a nodular polypoidal mass lesion seen involving ascending colon & extending upto hepatic flexure showing moderate to avid enhancement on arterial phase (Fig. 1), persistent contrast enhancement on portal phase & contrast wash out on delayed phase with multiple enlarged pericolic lymph nodes (Fig. 2). Colonoscopy was performed which revealed a polypoid nodular growth in ascending colon proximity to hepatic

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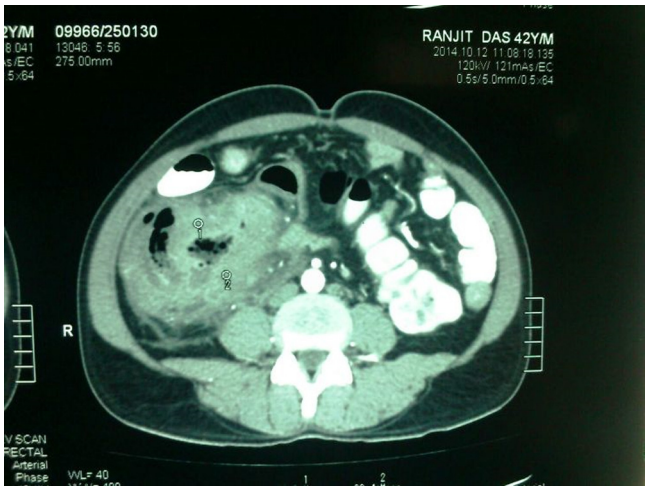

Fig. 1. CECT of whole abdomen showing an irregular bowel wall thickening (15–19 mm) with a nodular polypoidal mass lesion.

flexure with narrowed lumen and the scope was not negotiable further (Fig. 3). Multiple biopsies were taken which gave non-specific inflammatory lesion. So, inflammatory mass of colon was suspected, but clinically, carcinoma of ascending colon could not be excluded and was planned for surgery.

Right hemicolectomy with an end to end ileocolic anastomosis was done and a mass lesion in the ascending colon, near to hepatic flexure not associated with any enlarged lymph nodes seen. Histopathology of the specimen showed patchy destruction of colonic mucosa with dense infiltration of acute inflammatory cells in the ulcerated area, multiple foci of abscess comprising of polymorphs and nuclear debris in different layers of the wall starting from mucosa to serosa, presence of epithelial histiocytes and multinucleated giant cells, clumps as well as discrete fungal hyphae within these inflammatory areas, infrequently septate, uneven wide hyphae having wide angle branching in these clusters on colonic mass sections (Fig. 4). Serosal nodule section showed

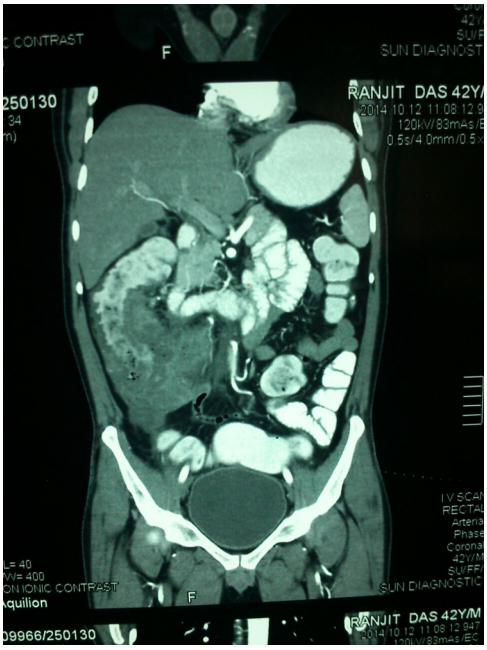

Fig. 2. CECT of whole abdomen showing an irregular bowel wall thickening with a nodular polypoidal mass lesion seen involving ascending colon & extending upto hepatic flexure with multiple enlarged pericolic lymph nodes.

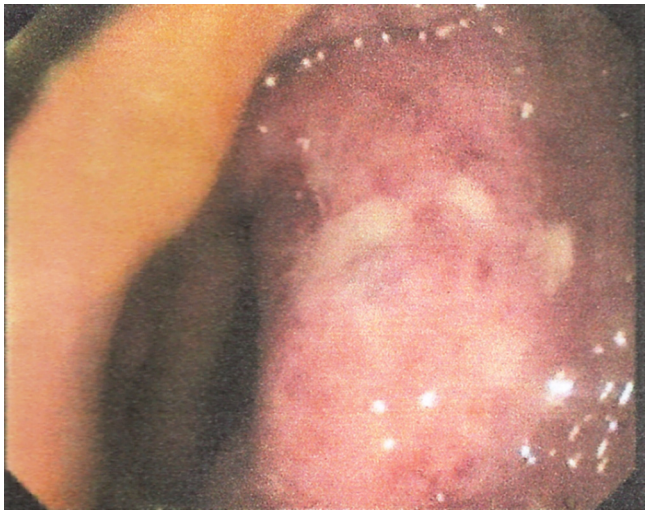

Fig. 3. Colonoscopic view of ascending colon proximity to hepatic flexure with a polypoid nodular growth with narrowed lumen.

necroinflammatory changes as described above with clumps of fungal hyphae and area of angioinvasion & thrombus formation (Fig. 5).

Upon receiving the pathology report, systemic intravenous amphotericin B treatment was initiated as colonic mucormycosis was confirmed on histopathology. Patient recovered well and was discharged on 14th postoperative day.

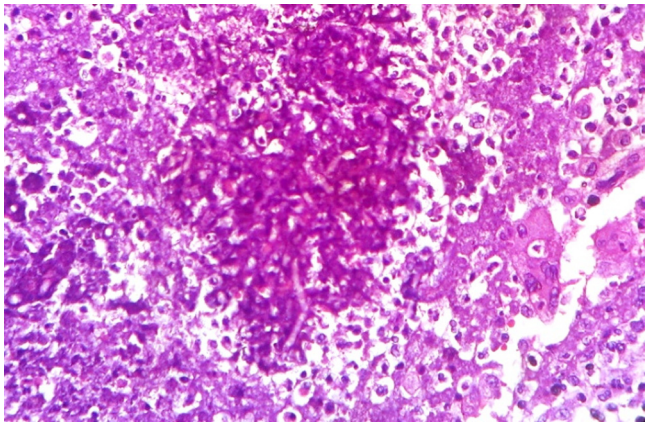

Fig. 4. Histopathology showing presence of epithelial histiocytes, multinucleated giant cells, clumps as well as discrete fungal hyphae within an inflammatory area having wide angle branching.

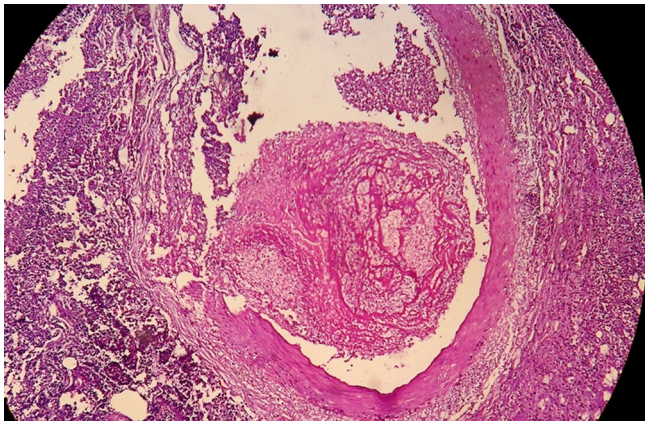

Fig. 5. Serosal nodule section showing area of angioinvasion & thrombus formation.

3. Discussion

Mucormycosis is a rare, opportunistic infection caused by the fungi from the class Zygomycetes, order mucorales. The fungus exists in two forms, the infective sporangiospores and the hyphal form, responsible for tissue necrosis and dissemination. Depending on the site of involvement, mucormycosis can be classified as rhinocerebral, pulmonary, cutaneous, GI, central nervous system or disseminated mucormycosis [3].

GI mucormycosis is the rarest of all forms of mucormycosis, accounting for approximately 7% of all cases [10]. GI mucormycosis most commonly involves the stomach (57.5%), followed by the colon (32.3%) and the ileum (6.9%). This rare and opportunistic infection has been reported in neonates, probably due to their immature immunity, presenting as necrotizing enterocolitis. Fewer than 20 cases of GI mucormycosis with colonic involvement have been reported in the past two decades [11]. GI infection most probably results from ingestion of fungal spores. Being an opportunistic infection, some degree of immunocompromised status is almost always associated with GI mucormycosis. The most common underlying risk factors for mucormycosis include poorly controlled diabetes mellitus, administration of high-dose systemic corticosteroids in solid organ and hematopoietic stem cell transplantation, penetrating trauma or burns, persistent neutropenia and deferoxamine-based therapy [12]. In our case, patient is a known case of type-II diabetes mellitus for last 10 years and he was not taking any antidiabetic medications since 2 years, resulting in uncontrolled diabetes.

A typical gastrointestinal lesion consists of a dark ulcer with sharply demarcated edges and with necrosis and thrombosis in adjacent vessels. The infection can extend from the lumen of the gut and may cause obstruction, perforation or bleeding. Initial presentations may be abdominal pain and distension, fever, and diarrhoea. If there is extensive bowel involvement with multiple ulcers caused by the fungal infection, it may present with gastrointestinal bleeding or even visceral perforation at late presentation [10]. Even though melena is a feature of upper GI bleeding, because we failed to demonstrate any specific lesion causing an upper GI bleed, it was presumed that colonic mucormycosis was responsible for melena in this patient.

Lack of pathognomic clinical features renders early diagnosis of GI mucormycosis very difficult. Although a vigilant clinician can suspect the possibility of GI mucormycosis in a patient with traditional risk factors, the same cannot be expected in a patient without traditional risk factors. The diagnosis of mucormycosis is rarely suspected and antemortem diagnosis is made in only 25–50% of cases [13]. Colonic mucormycosis presenting as a mass with altered bowel habit, melena and abdominal pain is extremely difficult to differentiate it from carcinoma colon. A definitive diagnosis of mucormycosis is almost always ascertained by histopathological evidence of fungal invasion of tissue. On CECT of whole abdomen, an irregular bowel wall thickening with a nodular polypoidal mass lesion, involving ascending colon & extending upto hepatic flexure with multiple enlarged pericolic lymphnodes seen which gave high suspicion of malignancy. In colonoscopy also we found same nodular polypoidal growth obstructing the lumen of ascending colon near hepatic flexure but colonoscopic biopsy showed nonspecific inflammatory lesion. Diagnosis depends on histological examination for the presence of predominantly aseptate wide hyphae with focal bulbous and non dichotomous branching occasionally at right angles [14]. Over 94% of sampled tissues also show infraction and angioinvasion on histology examination [15] as in our case.

Four factors are critical for eradicating mucormycosis: rapidity of diagnosis, reversal of the underlying predisposing factors (if possible), appropriate surgical debridement of infected tissue, and appropriate antifungal therapy. Early diagnosis is important because small, focal lesions can often be surgically excised before they progress to involve critical structures or disseminate [16]. Unfortunately, there are no serologic or PCR-based tests to allow rapid diagnosis. As mentioned, autopsy series have reported that up to half the cases of mucormycosis are diagnosed postmortem [17–19], underscoring the critical need to maintain a high index of clinical suspicion and to aggressively pursue diagnostic biopsy. Correcting or controlling predisposing problems is also essential for improving the treatment outcome. In diabetic ketoacidotic patients, hyperglycemia and acidemia should be corrected.

There are no recommendations regarding treatment specific to gastrointestinal infection. We managed our patient with right hemicolectomy and intravenous high dose (1 mg/kg per day) amphotericin B started after histopathological confirmation of colonic mucormycosis.

4. Conclusion

Mucormycosis is an increasingly common infection in immunocompromised patients. Knowing these unusual presentations of this disease, surgeon need to maintain a high index of suspicion and perform timely and appropriate diagnostic evaluation to improve patient outcome. Prompt diagnosis, reversal of predisposing conditions, and aggressive surgical debridement remain cornerstones of therapy for this deadly disease.

Conflict of interest

No.

Funding

No.

Consent

Yes.

Author Contribution

Dr. Prasanna K Debata: study design, data analysis.
Dr. Sangram Keshari Panda: study design, data analysis, writing.
Dr. Atmaranjan Dash: data collection, writing.
Dr. Ramakant Mohanty: study design, data analysis.
Dr. Biranchi N Mallick: study design, data analysis.
Dr. Debabrata Tadu: data collection, writing.
Dr. Vivek g nath: data collection, writing.
Dr. Abhinash Sahoo: data collection, writing.

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