CASE REPORT OPEN ACCESS

Gastrointestinal Mucormycosis in an Immunocompetent Patient: A Diagnostic Dilemma

Ramsha Ali[1,*], Abdul Rehman Alvi[2], Ursala Adil[3]

[1]*Dr Ruth KM Pfau Civil Hospital, Karachi, Pakistan*

[2]*Department of Surgery,* [3]*Aga Khan University Hospital, Karachi, Pakistan*

ARTICLE HISTORY

Received: Aug 28, 2024
Revised: Dec 28, 2024
Accepted: Dec 30, 2024

Citation: Ali R, Rehman A, Adil U. Gastrointestinal mucormycosis in an immunocompetent patient: a diagnostic dilemma. Acad Res. 2024; 1(2): 83-87.

DOI: https://doi.org/10.70349/ar.v1i2.1

Abstract

Mucormycosis is a rare fungal infection caused by mucorales. It mainly affects immunocompromised individuals. Mucormycosis can infect the gastrointestinal tract and the pathophysiology involves angioinvasion and necrosis of tissues.

We report a 45 year old immunocompetent patient presenting with complaints of abdominal pain and constipation with left upper quadrant tenderness on examination. He was treated along the lines of intra-abdominal abscess but as his condition deteriorated, he underwent Exploratory laparotomy which showed ischemic bowel segments. Histopathology examination of tissue samples revealed non septate branching fungal hyphae consistent with invasive mucormycosis. Antifungal therapy was started and the patient underwent relook surgery, extensive bowel necrosis was found all looking non salvageable and the surgery was abandoned.

This case report highlights the importance of keeping fungal aetiology in differentials of immunocompetent patients presenting with non-specific gastrointestinal symptoms with characteristic CT scan findings not responding to conventional antibiotic treatment. In addition, it is important to start prophylactic broad spectrum antifungals such as amphotericin B along with surgical interventions to prevent further systemic spread of the disease as delay in diagnosis can worsen the outcomes.

Keywords: Gastrointestinal, invasive mucormycosis, immunocompetent patients.

1. INTRODUCTION

Mucormycosis is a rare but serious fungal infection that can be fatal. It is caused by a group of fungi belonging to class zygomycetes and order mucorales, which are found in our environment [1]. Mucormycosis has become a significant concern, ranking third among invasive fungal infections in patients undergoing stem cell transplants, after candidiasis and aspergillosis. Additionally, it poses a threat to diabetic patients in Western countries. The disease is also increasingly detected in developing nations like India, particularly among individuals with uncontrolled diabetes or trauma. However, due to limited epidemiological data, understanding the full impact of mucormycosis remains challenging [2]. While mucormycosis can affect any organ system, the most common sites of infection are the sinuses (39%), lungs (24%) and skin (19%) [3, 4]. Although it is rare, mucormycosis can affect the gastrointestinal tract. This occurs when a person ingests fungal spores. An analysis of 176 cases, comprising both immunocompetent and immunosuppressed individuals, revealed that the majority (64.1%) of infections occurred in the intestinal tract, with a significant proportion (33%) affecting the stomach. Furthermore, a review of 89 adult cases showed a male-to-female incidence ratio of approximately 2:1, with an average age of 47.7 years [5]. Cases of Gastrointestinal mucormycosis in people with a healthy immune system are rarely reported, but the mortality rate can be as high as 85% [4]. This case report highlights the importance of keeping gastrointestinal mucormycosis as a differential diagnosis in patients presenting with non-specific GI symptoms and characteristic radiologic findings, as these measures can lead to good prognosis.

* Address correspondence to this author at the Dr Ruth KM Pfau Civil Hospital, Karachi, Pakistan; E-mail: ramshaali110@gmail.com

2. CASE REPORT

This was a 45-year-old male, presented to the emergency department of Aga Khan hospital with fever, left upper quadrant pain, nausea, constipation and abdominal distension for 10 days. Patient had a history of smoking and chronic alcohol use. He was a known case of diabetes mellitus. Physical examination showed a fully conscious patient, alert and oriented, his vital signs were; blood pressure of 156/95 mm hg, Pulse rate of 97 beats per minute, temperature 38.2’ C and respiratory rate of 18 breaths per minute. On abdominal examination there was a hard tender mass of 10 x 10 cm felt at the left upper quadrant, abdomen was distended with sluggish gut sounds. His cardiovascular and respiratory examinations were unremarkable. Outpatient

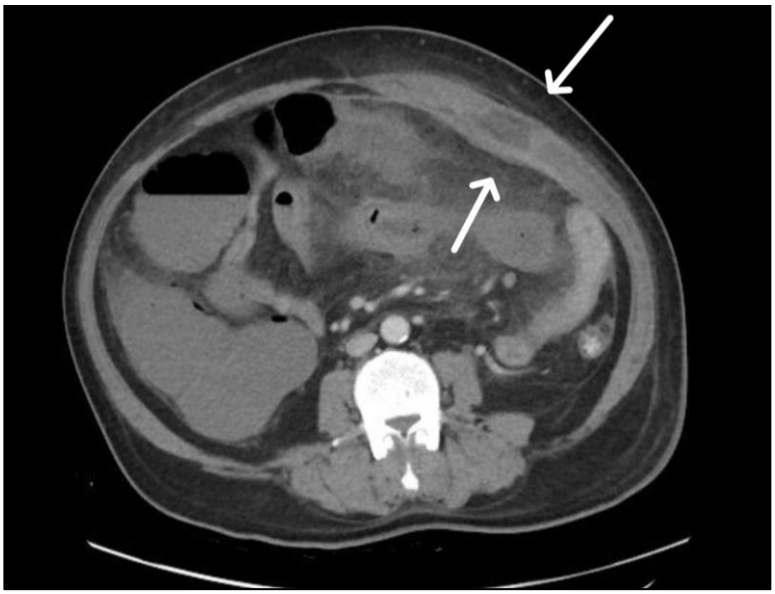

Figure 1: CT scan of abdomen showing left anterolateral abdominal wall thickening with fluid levels in the Intramuscular plane surrounding inflammation and non-enhancing areas.

CT scan of abdomen showed extensive mesenteric fat stranding along the distal part of transverse colon, thickening of left anterolateral abdominal wall with central hypodense area representing an abscess (Fig. **1-2**). Patient was resuscitated and treated along the lines of intra abdominal abscess with broad spectrum antibiotics and antiprotozoals. Patient was kept nil by mouth and NG tube was inserted. Due to persistence of his symptoms and no response to the given treatment CT scan was repeated which showed thickened long segment of transverse colon measuring 10 cm in length, non enhancing 12 cm segment of mid jejunum with significant surrounding inflammatory changes representing bowel ischemia. Left antero lateral abdominal wall inflammation and fluid tracking in intramuscular planes measuring 27x44x25 mm with 39 cc volume.

Emergency exploratory laparotomy was performed on second day of his admission, extensive omental infarction were seen covering all over distal transverse colon creating a phlegmon, distal transverse colon and jejunal ischemia

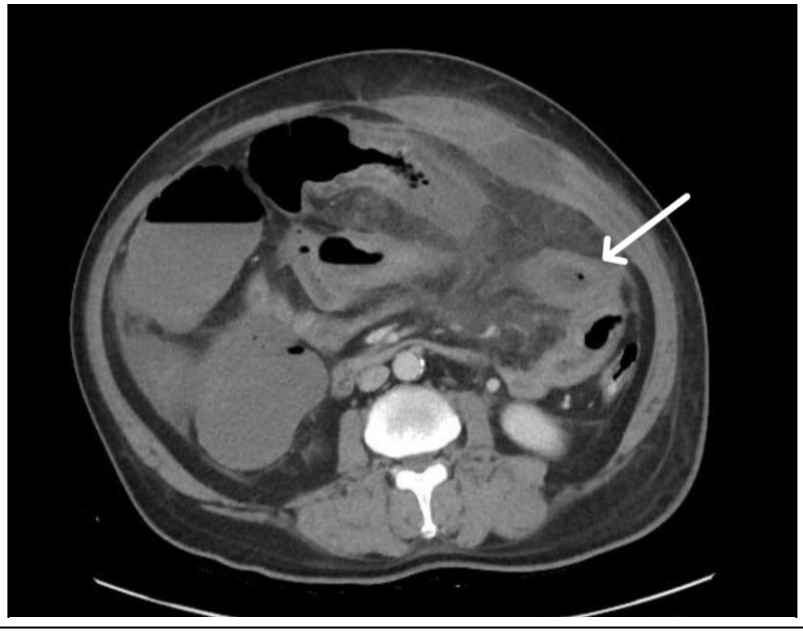

Figure 2: CT scan of abdomen showing thickened segment of large and small bowel loops on left side along with non-enhancement and surrounding inflammation with mesenteric involvement and fat stranding.

were present with corresponding mesenteric infarcts. Ischemic bowel segments were resected with formation of venting jejunostomy and colostomy, left anterolateral abdominal wall was debrided and pus was drained, samples were sent for histopathology and culture, abdomen was temporarily closed with bogota bag and patient was shifted to surgical ICU after damage control surgery. Patient was re-explored after 24 hours, intraoperative findings revealed necrotic patches on left anterolateral abdominal wall for which extensive debridement was performed, due to the clinical condition of the patient definitive surgery couldn't be performed and patient was shifted back to surgical ICU with bagota in place. Postoperatively the patient received inotropic support as he became hypotensive and vitally unstable. Provisional histopathology report revealed bowel infarction with mixed acute and chronic inflammation, thick walled congested mesenteric vessels having fibrin thrombi, exhibiting non-septate branching fungal hyphae and spores consistent with invasive mucormycosis, antifungals amphotericin B was started. However, the patient's clinical condition deteriorated, he developed metabolic acidosis and lactate remained high. After 48 hours of first surgery, the patient was taken for relook surgery and this time the necrotic area extended further from left side to right side involving muscles, fat with extensive necrosis of bowel loops all looking non salvageable with purulent ascites. Surgery was abandoned. The patient's family was counselled and they opted to change his code to DNR pharma followed by withdrawal of support. Following withdrawal, the patient became hypotensive and expired the following day due to cardiac arrest.

3. DISCUSSION

Gastrointestinal Mucormycosis is a severe life-threatening fungal infection caused by Mucorales species, it mainly affect immunocompromised individuals, there are only few cases of life threatening mucormycosis reported in immunocompetent hosts [3, 4] in our patient the only risk factor identified was chronic alcohol ingestion and diet controlled diabetes mellitus. In a study of 929 cases by Roden et al, GI manifestations were reported in only 7% of cases with mucormycosis [4] and the most frequently affected organs are the stomach (58%), followed by the colon (32%), small intestine (7%), and oesophagus (7%) [6] However, A review of 31 cases of gastrointestinal mucormycosis in immunocompromised patients revealed a notable trend: over the past two decades, there has been a rising incidence of mucormycosis affecting the intestinal tract [7]. Mucormycosis causes infarction and necrosis of host tissue by angioinvasion through fungal hyphae, gastrointestinal mucormycosis often presents with non-specific symptoms such as abdominal pain localised to affected area of gastrointestinal tract, distention, vomiting and constipation [4], similar symptoms were present in our patient. Patients diagnosed with intestinal mucormycosis consistently present with symptoms of fever, ranging from low to high grade, accompanied by abdominal pain. Interestingly, some cases have been discovered incidentally, where patients were initially hospitalized for unrelated underlying conditions, only to be later diagnosed with intestinal mucormycosis [8]. A notable case report described an unexpected diagnosis of colonic mucormycosis in a patient who was initially hospitalized for severe symptoms, including high fever, shock, and respiratory failure. Further investigation revealed that the patient had Streptococcus infantarius endocarditis, which had originated from a bacterial translocation in the colon [9] Gastrointestinal mucormycosis is a rare and fatal disease with rapid deterioration of patients and close to 50% of cases are diagnosed antemortem mainly by histopathologic examination of infected/necrotic tissues during surgery or endoscopy as cultures are usually negative [4,10, 11]. Currently, tissue diagnosis is the most reliable method for confirmation, and the use of Mucorales polymerase chain reaction (PCR) on formalin-fixed paraffin-embedded tissue enhances diagnostic accuracy. Microscopic examination of tissue reveals characteristic features of Mucorales, including broad (3-25 µm in diameter), thin-walled, and aseptate hyphae that appear folded, twisted, or compressed with irregular branching [12, 13]. The histopathology report in our patient reported similar findings of non-septate branching fungal hyphae and spores consistent with invasive mucormycosis. CT scan findings of small bowel mucormycosis are diffuse dilatation of the bowel with areas of wall thickening and decreased enhancement, colonic involvement presents as diffuse wall thickening associated pericolonic stranding and pneumatosis in thickened segments. This occurs due to varying degrees of arterial occlusion with or without reperfusion and/or venous occlusion [14], CT scan findings reported in our patient were thickened transverse colon and non enhancing segments of mid jejunum with surrounding inflammatory changes and loss of arterial arcade enhancement suggesting of bowel ischemia.

Effective management of gastrointestinal mucormycosis involves a multifaceted approach. First, it's essential to address the underlying conditions that increase the risk of infection, such as prolonged steroid use, co-morbidities, metabolic acidosis, and immunosuppression. Additionally, treatment requires a combination of surgical debridement of the infected tissue and antifungal therapy to combat the infection [15].

Mucormycosis can be treated medically and/or surgically, medical management alone is effective if early diagnosis is made and includes broad spectrum antifungal such as amphotericin B [16].

Due to the rarity of gastrointestinal mucormycosis, there is a lack of randomized controlled trials to evaluate the effectiveness of different antifungal treatments for this condition [17]. Timely management of mucormycosis is essential for better outcomes as delay of more than 6 days increases 30 day mortality from 35 to 66% [17]. This is supported by a retrospective study of 70 patients with hematological malignancies and concurrent mucormycosis

found that delayed or inadequate antifungal treatment significantly impacted prognosis. Specifically, patients who received antifungal therapy more than six days after diagnosis had nearly twice the mortality rate at 12 weeks compared to those who received timely and appropriate treatment [18]. In our patient surgical management was opted due to lack of histopathological diagnosis and no response to prophylactic antibiotics. Damage control exploratory laparotomy was done with removal of two necrotic bowel segments. This condition was difficult to manage due to late presentation of the patient after 7 days of symptoms with nonspecific complaints and tender left upper quadrant mistakenly thought to be an intra-abdominal abscess. In our patient antifungal treatment was initiated on the basis of provisional histopathology report. Hence in patients with non-specific gastrointestinal symptoms, it is essential to keep fungal aetiology in differentials if the patient does not respond to prophylactic antibiotics and start prophylactic broad spectrum antifungals to prevent further systemic spread of the disease as delay in diagnosis can worsen the outcomes and delay of more than 6 days can increase 30 day mortality from 35 to 66% [17] as seen in case of our patient who presented late. A case series of 66 reported cases gastrointestinal mucormycosis found a disturbingly high mortality rate, with 85% of patients with gastrointestinal involvement succumbing to the disease. The primary cause of these fatalities was bowel perforation, highlighting the severe nature of this infection [4].

CONFLICT OF INTEREST

The authors declare that none of them has a conflict of interest regarding the publication of this paper.

FUNDING

The study received no financial support.

ETHICAL APPROVAL AND PATIENT'S CONSENT

This case report adhered to the principles outlined in the Helsinki Declaration. However, formal IRB approval was not required as this is a single patient case report and it does not involve systematic research.

Written consent was obtained from the patient's immediate kin, for publication of this case report.

ACKNOWLEDGEMENTS

None.

AUTHOR'S CONTRIBUTION

RA: Data collection, literature review and drafting of the manuscript.
ARA: Conceived the idea and conceptualized the case report.
UA: Data collection and literature review.

.

REFERENCES

[1] Adhikari S, Gautam AR, Paudyal B, Sigdel KR, Basnyat B. Case report: gastric mucormycosis- a rare but important differential diagnosis of upper gastrointestinal bleeding in an area of Helicobacter pylori endemicity. Wellcome Open Res 2019;4:5. https://doi.org/10.12688/wellcomeopenres.15026.2

[2] Petrikkos G, Skiada A, Lortholary O, Roilides E, Walsh TJ, Kontoyiannis DP. Epidemiology and Clinical Manifestations of Mucormycosis, Clinical Infectious Diseases. 2012; 54(1): S23-S34. https://doi.org/10.1093/cid/cir866

[3] Maertens JH, Demuynck EK, Zache'e VP, Vandenberghe GEVP, Boogaerts MA. Mucormycosis in allogeneic bone marrow transplant recipients: report of five cases and review of the role of iron overload in the pathogenesis. Bone Marrow Transplant. 1999; 24: 307-12. https://doi.org/10.1038/sj.bmt.1701885

[4] Roden M, Zaoutis T. Epidemiology and outcome of zygomycosis: a review of 929 reported cases. Clin Infect Dis. 2005; 41(5):634-53. https://doi.org/10.1086/432579

[5] Kaur H, Ghosh A, Rudramurthy SM, Chakrabarti A. Gastrointestinal mucormycosis in apparently immunocompetent hosts-A review. Mycoses. 2018; 61(12): 898-908. https://doi.org/10.1111/myc.12798

[6] Bruno F, Barros B, Márcio LD, Lucas RS, Angelo S, Felipe NF. A rare case of gastric mucormycosis in an immunocompetent patient. Rev Soc Bras Med Trop. 2018; 51(3): 401-2. https://doi.org/10.1590/0037-8682-0304-2017

[7] Dioverti MV, Cawcutt KA, Abidi M, Sohail MR, Walker RC, Osmon DR. Gastrointestinal mucormycosis in immunocompromised hosts. Mycoses. 2015; 58(12): 714-8. https://doi.org/10.1111/myc.12419

[8] Antony SJ, Parikh MS, Ramirez R, Applebaum B, Friedman G, Do J. Gastrointestinal mucormycosis resulting in a catastrophic outcome in an immunocompetent patient. Infect Dis Rep. 2015; 7(3): 6031. https://doi.org/10.4081/idr.2015.6031

[9] Shah C, Zimmerman S, McKinney J, Ebers A. Colonic mucormycosis in an immunocompetent patient with endocarditis. IDCases. 2020; 20: e00773. https://doi.org/10.1016/j.idcr.2020.e00773

[10] Ribes JA, Vanover-Sams CL, Baker DJ. Zygomycetes in human disease. Clin Microbiol Rev. 2000; 13: 236-301. https://doi.org/10.1128/CMR.13.2.236

[11] Petrikkos G, Skiada A, Lortholary O, Roilides E, Walsh TJ, Kontoyiannis DP. Epidemiology and clinical manifestations of Mucormycosis. Clin Infect Dis. 2012; 54: 23-34. https://doi.org/10.1093/cid/cir866

[12] Hammond SP, Bialek R, Milner DA, Petschnigg EM, Baden LR, Marty FM. Molecular methods to improve diagnosis and identification of mucormycosis. J Clin Microbiol. 2011; 49(6): 2151-3. https://doi.org/10.1128/JCM.00256-11

[13] A colour atlas and textbook of the histopathology of mycotic disease. Postgrad Med J. 1981; 57: 471. https://doi.org/10.1136/pgmj.57.669.471

[14] Ghuman SS, Sindhu P, Buxi TBS, Sheth S, Yadav A, Rawat KS, Sud S. CT appearance of gastrointestinal tract mucormycosis. Abdom Radiol (NY). 2021; 46(5): 1837-45. https://doi.org/10.1007/s00261-020-02854-3

[15] Spellberg B, Walsh TJ, Kontoyiannis DP, Edwards J, Jr., Ibrahim AS. Recent advances in the management of mucormycosis: from bench to bedside. Clin Infect Dis. 2009; 48(12): 1743-51. https://doi.org/10.1086/599105

[16] Cornely OA, Alastruey-Izquierdo A, Arenz D, Chen SCA, Dannaoui E, Hochhegger B, et al. Global guideline for the diagnosis and management of mucormycosis: an initiative of the European Confederation of Medical Mycology in cooperation with the Mycoses Study Group Education and Research Consortium. Lancet Infect Dis. 2019; 19(12): e405-e421. https://doi.org/10.1016/S1473-3099(19)30312-3

[17] Werthman-Ehrenreich A. Mucormycosis with orbital compartment syndrome in a patient with COVID-19. Am J Emerg Med. 2021; 42: 264.e5-264.e8. https://doi.org/10.1016/j.ajem.2020.09.032

[18] Chamilos G, Lewis RE, Kontoyiannis DP. Delaying amphotericin B-based frontline therapy significantly increases mortality among patients with hematologic malignancy who have zygomycosis. Clin Infect Dis. 2008; 47(4): 503-9. https://doi.org/10.1086/590004