IDCases 31 (2023) e01709

Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcases

Case report

# A rare case of fatal gastrointestinal basidiobolomycosis

Mohammed Alsaeed [a,e,*,1], Mohamed Mursi [a], Abdelkarim Bahloul [a], Assem Alrumeh [b], Nahlah Arab [c], Mohannad Alrasheed [d]

[a] Medicine Department, Infectious Disease Division, Prince Sultan Military Medical City, Riyadh, Saudi Arabia
[b] Pathology Department, Prince Sultan Military Medical City, Riyadh, Saudi Arabia
[c] General Surgery Department, Colorectal Surgery Division, Prince Sultan Military Medical City, Riyadh, Saudi Arabia
[d] Radiology Department, Prince Sultan Military Medical City, Riyadh, Saudi Arabia
[e] Alfaisal University College of Medicine, Riyadh, Saudi Arabia

ARTICLE INFO

*Keywords:*
Basidiobolomycosis
Gastrointestinal Basidiobolomycosis
Eosinophilic colitis
Splendore–hoeppli phenomenon

ABSTRACT

Basidiobolomycosis is an uncommon fungal infection caused by the environmental saprophyte *Basidiobolus ranarum*. Basidiobolomycosis typically manifests as a subcutaneous infection, and rarely affects the gastrointestinal tract. It lacks a distinct clinical manifestation, and most initial cases are incorrectly identified. We report a 69-year-old male patient who presented to the emergency department with history of abdominal pain, fever, and weight loss for 1 year that turned to be gastrointestinal basidiobolomycosis.

## Introduction

Gastrointestinal Basidiobolomycosis (GIB) is an uncommon fungal infection caused by the environmental saprophyte *Basidiobolus ranarum* [1]. The first case of GIB was reported in a Nigerian child in 1964 [2]. Most of reported cases are in immunocompetent children with no apparent risk factors. Majority of cases were reported from United States of America, Saudi Arabia, India, and Iran [3].

Basidiobolomycosis typically manifests as a subcutaneous infection, and rarely affects the gastrointestinal tract. The infection occurs after a scratch or puncture from an insect, plant, or other objects that carry fungi [4]. The gastrointestinal tract has also been found to become infected after ingestion of soil, animal feces, or food contaminated with *Basidiobolus ranarum*. It lacks a distinct clinical manifestation, and most initial cases are incorrectly identified [5]. Here we report a rare case of a fatal GIB accompanied by refractory bacterial intraabdominal septic shock.

## Case presentation

Here, we present a 69-year-old male patient who resides in southern Saudi Arabia and works as a farmer. His past medical history includes uncontrolled type II diabetes and laparoscopic cholecystectomy one year prior to presentation. He presents to the emergency department on (day 0), with a two-week history of left lower abdomen pain, a one-year history of intermittent fever, and weight loss.

Initially, abdominal examination revealed paraumbilical and left lower quadrant tenderness with distension but no organomegaly and other systems where unremarkable.

Laboratory investigations showed leucocytosis 22.95 * 10^9/L with a differential of neutrophilia and eosinophilia (21.7 %), Haemoglobin of 126 g/L,Platelets of 518 * 10^9/L,C-reactive protein 262.48 mg/L (rest of labs were normal). Abdominal x-ray showed faecal impaction, nonspecific distribution of bowel gases, no air under diaphragm and no signs of bowel obstruction. CT abdomen and pelvis with contrast showed multifocal segmental wall thickening in the distal ascending, proximal transverse, and distal descending colon, with fluid collections suggesting concealed perforations (day 0).

The patient was admitted to the general surgery unit with a diagnosis of severe diverticulitis, and he was started on Piperacillin/Tazobactam and maintained nothing by mouth. The patient's symptoms worsened, and he had blood-stained stool. As a result, CT abdomen and pelvis with contrast was repeated, demonstrating recurrence of multifocal segmental wall thickening involving the hepatic flexure, proximal transverse, and distal descending colon, as well as evidence of concealed perforation and interval increase in size of surrounding collections (day 4). Overall findings were suggestive of severe diverticulitis versus aggressive infections like basidiobolomycosis and actinomycetes

* Correspondence to: Medicine department, Infectious disease division, Prince Sultan Military Medical City, P.O.Box 7897, Riyadh 11159, Saudi Arabia.
*E-mail address:* mohalsaeed@live.com (M. Alsaeed).
[1] ORCID: 0000-0002-0503-2154.

https://doi.org/10.1016/j.idcr.2023.e01709
Received 17 January 2023; Received in revised form 5 February 2023; Accepted 11 February 2023
Available online 13 February 2023

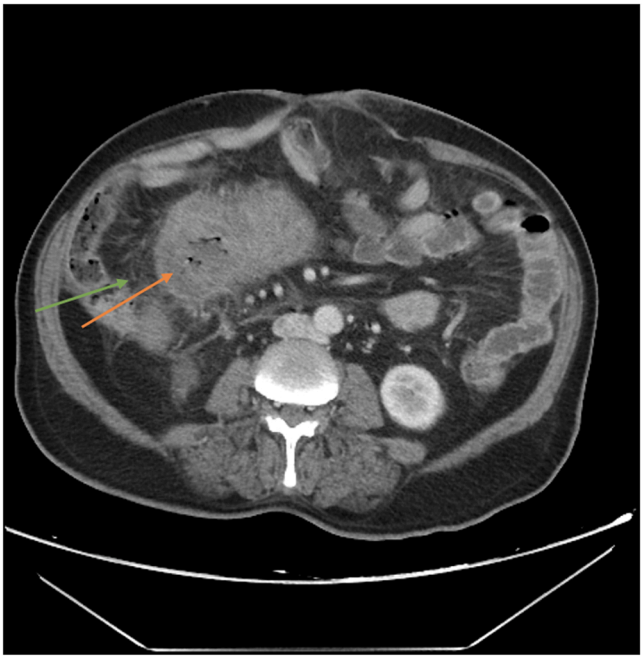

**Fig. 1.** Enhanced Axial CT scan of Abdomen & Pelvis showed thickening of the transverse colon (orange arrow) with peri-colonic fat stranding (green arrow).

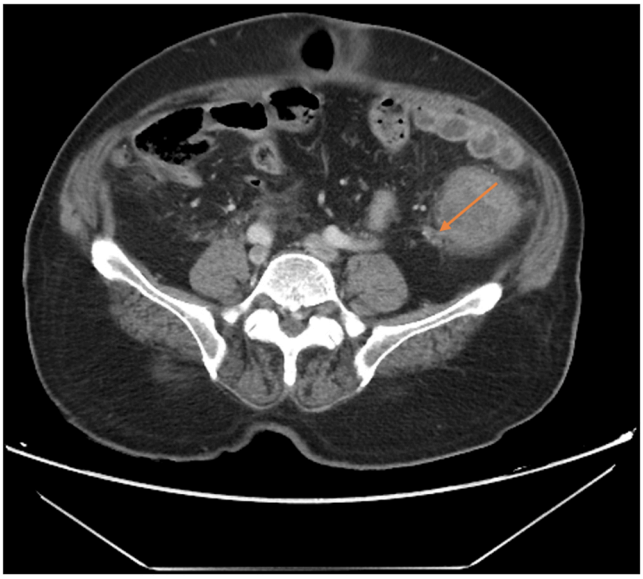

**Fig. 2.** Enhanced Axial CT scan of Abdomen & Pelvis showed thickening of the descending colon with sub-colonic abscess (orange arrow).

(Figs. 1–3).

Infectious diseases team was involved after release of the second abdominal CT result and recommended for surgical intervention, obtaining biopsy for bacterial, TB, fungal cultures, and histopathology in addition to an empiric start of liposomal amphotericin B, Amoxicillin/Clavulanic acid and tigecycline.

Although kept on broad spectrum antimicrobial agents, patient condition worsened, and he was persistently febrile. His white blood cells increased to 31.7 * 10^9/L with absolute eosinophils count increased to 3.58 * 10^9/L. His haemoglobin dropped to 10 g/dL therefore, a third abdominal CT with contrast was arranged that revealed redemonstration of circumferential wall thickening of the colon but interval increase in size of fluid collection measuring

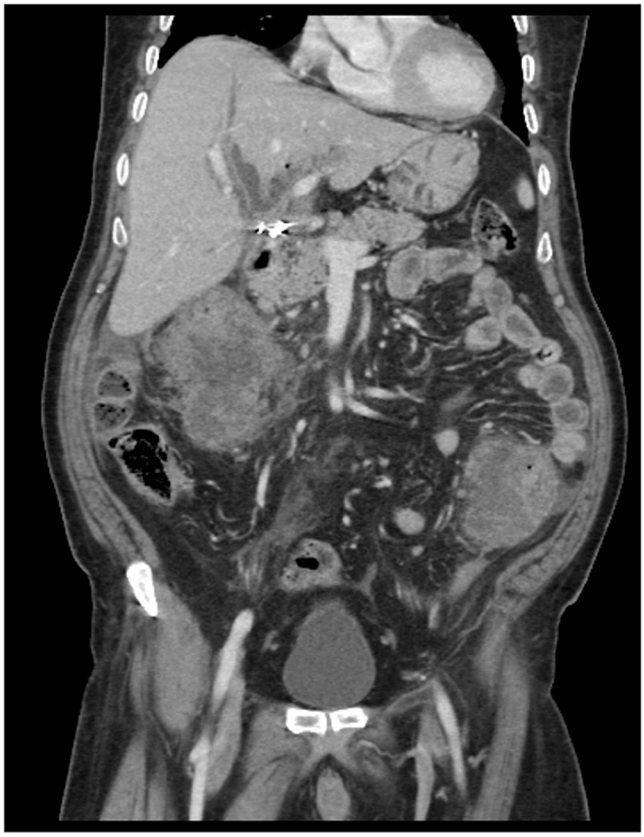

**Fig. 3.** Enhanced Axial CT scan of Abdomen & Pelvis showed severe thickening of the transverse and descending colon with peri-colonic abscess secondary to contained perforation.

approximately 7.9 × 9 cm, compared to 7.7 × 9.1 cm as well as interval increase in abdominopelvic ascites with surrounding inflammatory changes (day 8).

The patient was taken to operation room and found to have faecal peritonitis due to multiple colon perforation at sigmoid, transverse, and descending colon with small bowel adhesion and adhesions between small bowel and transverse colon. Adhesolysis was done in addition to total colectomy and end ileostomy was created with placement of two drains (day 9).

After the operation, the patient was transferred to intensive care unit (ICU) for close monitoring and observation. Interestingly, his absolute eosinophils count dropped immediately after surgery and reached down to 0.08 * 10^9/L.

While being in ICU, the patient's haemoglobin dropped to 69.0 g/L, and he was started to inotropic support and transfused several units of packed red blood cells. He maintained minimal ventilator settings with minimal respiratory secretions. Abdominal and pelvic CT was repeated two days after surgery and showed no collection found.

Bacterial tissue culture showed heavy growth of *Pseudomonas aeruginosa* and carbapenem resistant *Klebsiella variicola*. Blood culture from the central line, showed *Bacteroides fragilis*. Fungal tissue culture showed *Candida glabrata*. Histopathology showed extensive inflammation involving the full thickness of the colon tissue and extended to the serosal surface. The inflammation consists of mixed inflammatory cells along with sheets of eosinophils and multiple foci of fungal microorganism. The fungal microorganisms are hyphae that are irregularly branched, thin walled, occasionally septated and surrounded by thick eosinophilic cuff splendore-Hoeppli phenomenon (Figs. 4–6).

Despite surgical intervention, continuing liposomal amphotericin B 5 mg/kg IV once per day, Itraconazole 200 mg orally twice per day and modifying antibacterial agents; patient's condition deteriorated and

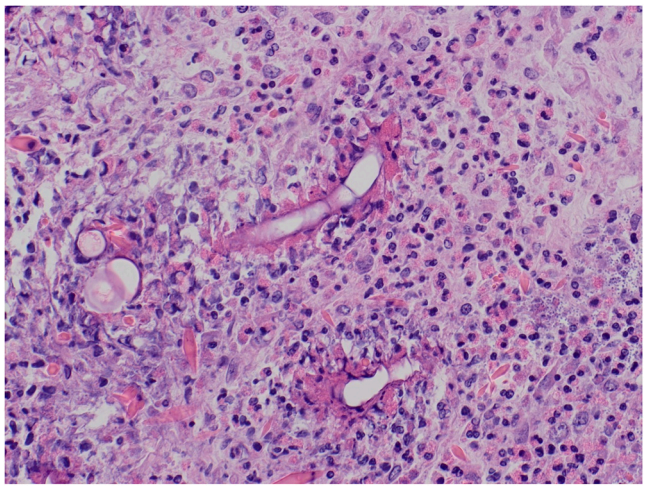

**Fig. 4.** H&S stain show thin wall and broad hyphae surrounded by eosinophilic material.

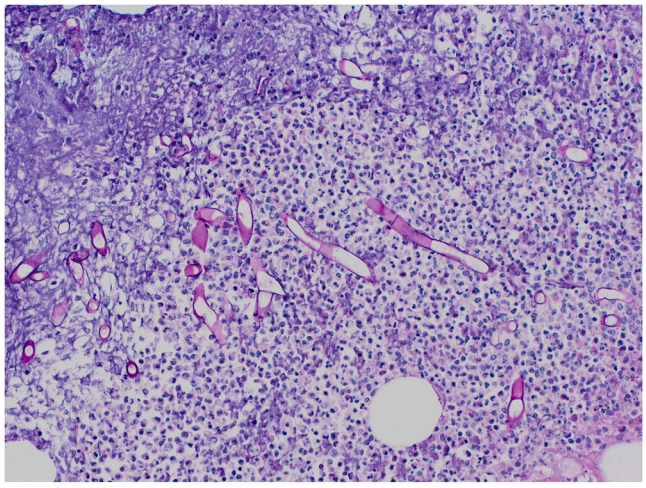

**Fig. 5.** Periodic acid-Schiff (PSA) stain highlights fungal wall.

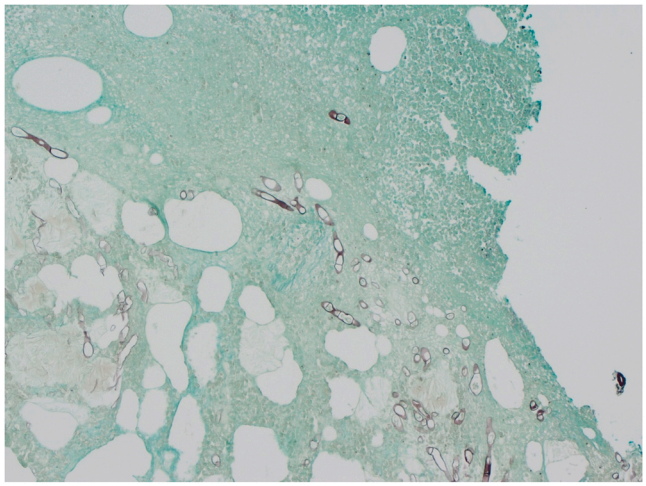

**Fig. 6.** Gomori Methenamine Silver (GMS) stain highlights fungal Wall.

passed away due to refractory septic shock.

## Discussion

Basidiobolus ranarum is a member of the Entomophthorales order, which was previously classified within the Zygomycetes class [6]. In immunocompetent individuals, the disease manifests primarily as a chronic subcutaneous non-angioinvasive infection. GIB is an uncommon, emerging, potentially fatal mycotic infection that primarily affects immunocompetent hosts. Approximately 100 cases of GIB have been reported to date. Typically observed in male pediatric patients [3].

GIB is difficult to diagnose clinically due to its nonspecific clinical manifestations, such as fever, abdominal pain, and abdominal mass. Chronic Infections of the GI tract, inflammatory bowel disease and tumors share similar presentation. Pezzani et al. reviewed 102 cases of GIB reported in the medical literature; abdominal pain (86 %) and fever (40 %) were the most frequently reported symptoms, and approximately one-third of the patients had abdominal masses on physical examination most commonly involving the colon. In 85 % of patients, the most notable laboratory investigation was peripheral eosinophilia [3]. In a similar review of GIB pediatric patients from Saudi Arabia, Shreef et al. found that abdominal pain and mass were also the most common presentation, with fever occurring in only 22 % of patients [7].

Culture is the gold standard for diagnosing condition definitively. Sabouraud agar is the medium used for isolation of Basidiobolus ranarum and the growth needs 2–3 days in 25–30 °C. Colonies appear as white to gray with radial folds in the plates [8]. Due to low yield of cultures, most reported cases like our patient are diagnosed based on histopathology. Typical morphological features include granulomatous inflammation and diffuse eosinophilic infiltrate with thin-walled branched hyphae surrounded by eosinophilic material (Splendore-Hoeppli phenomenon) [9]. Recent use of a molecular test to diagnose basidiobolomycosis has demonstrated its high sensitivity and specificity reaching 100 % [10].

The disease's rarity makes it difficult to recommend the most effective treatment. The majority of reported cases were treated surgically and medically. Few cases were successfully treated with only medical therapy. The experience with azole specifically prolonged course of itraconazole after surgery has been widely used [11]. Since resistance has been observed in over fifty percent of cases, amphotericin B is no longer appropriate [12]. The reported death rate for GIB is close to 20 %, with no obvious cause; this may be attributable to late presentation with its complications [3].

In conclusion, our patient presented from southern Saudi Arabia, where the majority of GIB cases have been reported. He had abdominal pain, fever and eosinophilia raising the possibility of GIB. His symptoms were chronic, and his hospital course progressed rapidly, despite early coverage with broad antimicrobials and surgical intervention he passed away with refractory intra-abdominal sepsis.

## Ethical approval

Not applicable.

## Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

## CRediT authorship contribution statement

Mohammed Alsaeed: writing the manuscript and taking care of the patient. Mohamed Mursi: writing the manuscript and taking care of the patient. Abdelkarim Bahloul: reviewing the manuscript and taking care of the patient. Assem Alrumeh: reviewing the manuscript and taking care of the patient. Nahlah Arab: reviewing the manuscript and taking

care of the patient. Mohannad Alrasheed: reviewing the manuscript and taking care of the patient.

## Conflict of Interest Statement

The authors have no conflicts of interest to disclose.

## Acknowledgments

None.

*Consent*

Available upon request.

## References

[1] Al-Shanafey S, AlRobean F, Bin Hussain I. Surgical management of gastrointestinal basidiobolomycosis in pediatric patients. J Pediatr Surg 2012;47:949–51.
[2] Edington GM. Phycomycosisinibadan, western Nigeria. Trans R Soc Trop Med Hyg. 58; 1964. p. 242–5.
[3] Pezzani MD, Di Cristo V, Parravicini C, Sonzogni A, Tonello C, Franzetti M, Sollima S, Corbellino M, Galli M, Milazzo L, Antinori S. Gastrointestinal basidiobolomycosis: an emerging mycosis difficult to diagnose but curable. Case report and review of the literature. Travel Med Infect Dis 2019;31:101378.
[4] Mugerwa JW. Subcutaneous phycomycosis in Uganda. Br J Dermatol 1976;94:539–44.
[5] Bittencourt AL, Ayala M, Ramos EAG. A new form of abdominal zygomycosis different from mucormycosis. Am J Trop Med Hyg 1979;28:564–9.
[6] Kwon-chung KJ. Taxonomy of fungi causing mucormycosis and Entomophthoramycosis (zygomycosis) and nomenclature of the disease. Clin Infect Dis 2012;54:S8–15.
[7] Shreef K, Saleem M, Saeedd M, Eissa M. Gastrointestinal basidiobolomycosis an emerging, and a confusing, disease in children(a multicenter experience). Eur J Pediatr Surg 2016;5(5):131–4.
[8] El-Shabrawi MH, Kamal NM, Kaerger K. VoigtK, Diagnosis of gastrointestinal basidiobolomycosis: a mini-review. Mycoses 2014;57:138–43.
[9] Almoosa Z, Alsuhaibani M, Aidandan S, Alshahrani D. Pediatric gastrointestinal basidiobolomycosis mimicking malignancy. Med Mycol Case Rep 2017;18:31–3.
[10] Gómez-Muñoz MT, Fernández-Barredo S, Martínez-Díaz RA, Pérez-Gracia MT, Ponce-Gordo F. Development of a specific polymerase chain reaction assay for the detection of Basidiobolus. Mycologia 2012;104(2):585–91.
[11] Al-Naemi AQ, Ali Khan L, Al-Naemi I, Amin K, Ali Athlawy Y, Awad A, et al. A case report of gastrointestinal basidiobolomycosis treated with voriconazole. A rare emerging entity. Medicine 2015;94:e1430.
[12] Yusuf NW, Assaf HM, Rotowa NA. Invasive gastrointestinal *Basidiobolus ranarum* infection in an immunocompetent child. Pediatr Infect Dis J 2003;22:281–2.