CASE REPORT

Basidiobolomycosis: a case of a rare fungal infection misdiagnosed as adenocarcinoma of the colon

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ABSTRACT

Background: Basidiobolomycosis is a rare infection caused by the fungus Basidiobolus ranarum. Since 1887, B. ranarum has been identified as an unusual fungus. It is a saprophytic fungus found in soil, decaying fruits and vegetables, and the digestive tracts of amphibians, fish, reptiles, and insectivorous bats. Originally, this group of fungi belonged to the Zygomycetes family, which has two orders including the Mucorales and the Entomophthorales, which display exceptional pathological manifestations. Only immunocompromised patients are affected by the Mucorales, whereas immune-competent individuals are affected by the Entomophthorales. Since its discovery in 1964, this unusual and rare fungal infection has only been reported 71 times in the literature, and few cases were reported in Saudi Arabia.

Case Presentation: A 62-year-old female Burmese with a 2-month history of nausea, vomiting, diarrhea, and chronic abdominal pain, accompanied by weight loss, loss of appetite, and lower limb myopathy was presented. This myopathy was so severe that it rendered her bedridden, thereby diminishing her quality of life. No hemoptysis or bleeding per rectum was seen. Past surgical and medical history were unremarkable. No medications or allergies were reported. Regarding her social history, she was married and a housewife with low socioeconomic status, and a significant language barrier. The patient was successfully diagnosed and properly treated.

Conclusion: Basidiobolomycosis is an emerging fungal pathogen. It usually causes subcutaneous infection, transmitted via traumatic inoculation. Gastrointestinal basidiobolomycosis is rare; it is associated with mass or inflammatory lesions in the gastrointestinal tract.

Keywords: Basidiobolomycosis, rare, fungal infection, colon, case report.

Introduction

Basidiobolomycosis is a rare infection caused by the fungus Basidiobolus ranarum. Since 1887, B. ranarum has been identified as an unusual fungus. It is a saprophytic fungus found in soil, decaying fruits and vegetables, and the digestive tracts of amphibians, fish, reptiles, and insectivorous bats. Originally, this group of fungi belonged to the Zygomycetes family, which has two orders including the Mucorales and the Entomophthorales, which display exceptional pathological manifestations. Only immunocompromised patients are affected by the Mucorales, whereas immune-competent individuals are affected by the Entomophthorales. Since its discovery in 1964, this unusual and rare fungal infection has only been reported 71 times in the literature and few cases were reported in Saudi Arabia.

Patients might exhibit abdominal pain, fever, constipation, and gastrointestinal bleeding, among other clinical manifestations. Due to the unfamiliarity of this infection, however, confusion arises among physicians, resulting in diagnostic delays.

Basidiobolomycosis of the gastrointestinal tract is frequently misdiagnosed as lymphoma or carcinoma, tuberculosis (TB), or inflammatory bowel disease. A case of gastrointestinal tract infection basidiobolomycosis (GIB) was presented which was initially misdiagnosed as cancer. The patient in the

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A 62-year-old female Burmese with a 2-month history of nausea, vomiting, diarrhea, and chronic abdominal pain, accompanied by weight loss, loss of appetite, and lower limb myopathy was presented. This myopathy was so severe that it rendered her bedridden, thereby diminishing her quality of life. No hemoptysis or bleeding per rectum was seen. Past surgical and medical history were unremarkable. No medications or allergies were reported. Regarding the social history, she was married and a housewife with low socioeconomic status, with a significant language barrier.

After requesting to be discharged, the patient was followed up in a private clinic and was diagnosed with adenocarcinoma of the colon based on a computerized tomography (CT) scan. No biopsies were taken. At the private clinic, everything was explained to the patient, and she was made fully oriented about the severity of her condition as the mass had an obstructive effect. Unfortunately, the patient denied the surgery. And ultimately, she was presented to the emergency department with septic shock.

On clinical examination, the patient was feverish and appeared sick, cachectic, and dehydrated. Vitaly, the blood pressure was 98/61, the heart rate was 136, and SPO2 was 96% on room air. No air under the diaphragm was noted (Figure 1).

The abdomen was rigid and distended, and a generalized tenderness which was going with peritonitis. There was a palpable mass extending from the pelvis to above the umbilicus. She was admitted under medical as a case of septic shock, dehydration, and pneumonia in the intensive care unit (ICU). The surgical department was involved as she had abdominal peritonitis.

Laboratory examinations were also performed (Table 1). CT scan with contrast of the abdomen showed the presence of significant free fluid, suggesting a pneumoperitoneum that might be caused by a perforated bowel (Figure 2).

A developing abscess was also noted. The scan also showed a generalized enhancement of the bowel mucosa that could raise the suspicion of a shocked bowel (Figure 3). Differential diagnosis includes malignant perforation, perforated viscus, inflammatory bowel, abdominal TB, lymphoma, and bowel ischemia. Upon presentation to the emergency department, the patient was in septic shock. Therefore, she was intubated, kept on in vitro fertilization, inotropic support started, and broad-spectrum antibiotics (tazocin, metronidazole, and vancomycin) were initiated. Due to her rapid deterioration, she was urgently transferred to the operating room for an exploratory laparotomy.

A massive mass with necrosis that extended from the hepatic flexure to the splenic flexure with multiple perforations and adhesions to the small bowel, pancreas, retroperitoneum, and lesser omentum was noted. Two additional masses were found. The first was about 20 cm from the ileocecal, and the other was in the middle of the left colon. A resection of 20 cm from the ileocecal, ascending, and transverse part of the splenic flexure was performed.

**Figure 1.** CXR of the patient’s abdomen showing no air under the diaphragm.

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<th><strong>Table 1. Patient’s laboratory findings (values in italics are abnormal).</strong></th>
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was done, and an ileostomy was created. Extended right hemicolectomy with ileostomy. Multiple biopsies were collected from the resected material for histopathologic investigations in order to reach a definitive diagnosis. The gross examination of the right hemicolectomy specimen showed an inflamed and markedly thickened wall, with multiple deposits. The mass-like thickened wall measures $20 \times 12 \times 12$ cm, and it is 10 cm away from the distal surgical margin. No fissures or fistulas were seen. The outer surface of the specimen was irregular, firm, and had a white-yellowish appearance. Occasional white patches were seen on the serosa of the colon. Upon opening the specimen, the ileal wall and transverse colon wall near the distal margin showed areas of soft tan—the appearance of mucosa with several yellow nodules involving mucosa and submucosa. The ileocecal valve, cecal wall, and proximal two-thirds of the ascending colon mucosa were unremarkable. There was a mass-like, marked hypertrophic circumferential thickening of the wall of the distal one-third of the ascending colon and two-thirds of the transverse colon with marked narrowing of the lumen and extensive ulceration (Figure 4).

The microscopic examination showed a cecal mass, transverse and longitudinal sections of broad fungal hyphae surrounded by the eosinophilic cuff, forming a sunburst pattern known as Splendore-Hoeppli phenomenon (asteroid bodies). H&E stain with a background of acute inflammatory cells and microabscess. Both resection margins were free, with patchy serositis. Three lymph nodes were identified, and all were free of fungal infection. No atypia or malignancy was seen.

The colonic wall showed marked ulceration of the distal ascending and transverse colon. Mucosal ulceration with a dense and marked transmural infiltration by eosinophils, neutrophils, and chronic granulomatous inflammation with central abscesses, multinucleated giant cells (foreign body type), and plasma cells, and fibrosis were noted. Fungal hyphae were noted invading the bowel wall with characteristic (Splendore-Hoeppli phenomenon) bodies demonstrating the fungus surrounded by eosinophilic material. The fungus was also surrounded by inflammatory cells, with some fungi engulfed by multinucleated giant cells. The overlying intact mucosa shows prominent eosinophils in the lamina propria. No cryptitis or crypt abscess was noted. Zones of coagulative-type necrosis were noted in the colonic wall. The inflammatory reaction reaches up to the pericolic fat with multiple fungal organisms detected in the pericolic fat. Massive areas of fat necrosis with bacterial colonies were noted. Focal multinucleated giant cells and fungal organisms were seen in the wall of blood vessels. A similar pathological process was seen in the terminal ileum, near the proximal surgical margin, and in the distal transverse colon near the distal surgical margin (Figure 5).
The patient was returned to the ICU post-operation. The patient was kept on endotracheal intubation until she reached a stable condition. She was kept on antibiotics in addition to anti-fungal treatment (voriconazole). Days later, she was extubated and transferred to the surgical ward.

The final diagnosis of colonic basidiobolomycosis was made. Further workup of the MRI spine showed lumbar spondylosis with disc bulge and herniation. L5 vertebral body and the right pedicle lesion might represent metastasis or spreading infection in the clinical setting of a previous abdominal mass (Figure 6).

Chest CT with contrast IV showed no evidence of mediastinal or hilar nodal enlargement. No detectable lesions in the chest wall were seen. Scans through the upper abdomen were normal. Impression of bilateral pleural effusion with collapsed lower lobes in both lungs was observed. Ground glass densities seen in both lungs suggested lung infection.

The patient spent almost one month in the hospital, seen by different specialties, including infectious disease, and they started her on Voriconazole 200 mg bid orally. She was discharged on anti-fungal Voriconazole 200 mg orally BID for six months, antibiotics (tazocin, metronidazole, and vancomycin), and multivitamins with ensure supplement drink. The patient was following in out patient departments for nearly 3 months, the laparotomy wound was clean, and the stoma was functionally well.

Discussion

Since 1887, *B. ranarum* has been identified as an unusual fungus. It is a saprophytic fungus found in soil, decaying fruits and vegetables, and the digestive tracts of amphibians, fish, reptiles, and insectivorous bats [1].

In 1956, a 4-year-old boy in Indonesia was diagnosed with Subcutaneous Mycosis, which was the very first case of Basidiobolomycosis to be documented [2]. At the time, physicians presumed that this opportunistic fungus caused only subcutaneous, nasal, and sinus infections.

Only a few years later, in 1964, reports of gastrointestinal basidiobolomycosis began to emerge, focusing on warm, humid tropical regions where the fungus thrived. The majority of cases involved Nigeria, East Africa, and West Africa, Iran, Saudi Arabia, and Kuwait [3].

The precise fungal source of infection remains unknown. Several logical hypotheses have been developed by researchers based on the available literature. The proposed theories included consuming food contaminated with reptile waste, soil, or animal feces; consuming putrefying

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**Figure 5.** Microscopic revelation showed focal multinucleated giant cells and fungal organisms in the wall of blood vessels.

**Figure 6.** MRI spine showed lumbar spondylosis with disc bulge and herniation.
Basidiobolomycosis

vegetables [4]; and directly infecting the perineum through "toilet leaves" that are used to clean the skin following a bowel movement.

The last hypothesis was supported by the observation that lesions were most noticed in the buttocks, thighs, and perineum [5]. An additional implausible theory involves implanted fungi after surgery, ranitidine consumption, smoking, and specific occupations such as gardening and landscaping [6].

People who have been diagnosed with GIB, which most commonly affects the colon, frequently experience abdominal pain, nausea, vomiting, diarrhea, or an abdominal mass. In the vast majority of cases, basidiobolomycosis of the gastrointestinal tract progresses slowly and locally, and it might be misdiagnosed as Crohn’s disease. In the cases of larger abdominal masses, it might also be mistaken for malignancy [5].

A limited number of cases worldwide, nonspecific clinical manifestation, and non-identifiable risk factors were all contributing factors that led to the misdiagnosis or underdiagnosis of GIB which then resulted in a consequent catastrophic complication [7]. Therefore, in order to reach maximum patient safety, GIB should be put in the differential diagnosis of any patient presenting with abdominal pain, fever, an abdominal mass, and significantly high ESR and eosinophilia [6].

Some studies reported that surgical intervention (laparotomy and resection of the inflamed part of the bowel) combined with medical therapy was a successful treatment of GIB. On the other hand, in some reported cases, surgical intervention was avoided and the patient was managed with prolonged medical therapy (Voriconazole). However, the need for surgical intervention is strictly dependent on the patient’s condition, the nature of the disease, and its location and extension.

After establishing a diagnosis of colonic and intestinal fungal infection, which is characteristic of the GIB, prompt therapy is required to eradicate the disease and prevent its early recurrence. Delays in treatment might lead to complications, including bowel perforation, obstructive uropathy, esophageal varices, and duodenal injury fistulas [6].

Numerous therapies have been used to eradicate this fungus, i.e., *B. ranarum*. Although it is not always successfully treated, the treatment typically involves potassium iodide (KI) and itraconazole. Other medications, such as trimethoprim-sulfamethoxazole, amphotericin B, and oral azoles such as ketoconazole (400 mg per day), have also been utilized. However, for the presented case, the most effective regimen consists of preoperative and postoperative itraconazole treatment for at least 6 months. Fortunately, no itraconazole-resistant fungi have been reported to date.

**Conclusion**

Basidiobolomycosis is an emerging fungal pathogen. It usually causes subcutaneous infection, transmitted via traumatic inoculation. Gastrointestinal basidiobolomycosis is rare; it is associated with mass or inflammatory lesions in the gastrointestinal tract. Cases have occurred worldwide, particularly in the Middle East and the southwestern part of the United States. Such patients are presented with abdominal pain and gastrointestinal symptoms; eosinophilia is typically 1,000 to 2,000/μl. The diagnosis is established via histopathology and culture. Surgery plus antifungal therapy was employed in the majority of patients. The most effective regimen consists of preoperative and postoperative itraconazole treatment for at least 6 months.

**List of Abbreviations**

CT Computerized tomography
ESR Erythrocyte sedimentation rate
ICU Intensive care unit
KI Potassium iodide
TB Tuberculosis

**Conflict of interest**
The authors declared that there is no conflict of interest regarding the publication of this case report.

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**Consent of publication**
Informed consent was obtained from the participant.

**Ethical approval**
Ethical approval is not required at our institution for an anonymous case report.

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**References**
