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# Gastrointestinal Basidiobolomycosis: Colonoscopy-Guided Biopsy and Effective Oral Antifungal Treatment

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## ABSTRACT

*Basidiobolus ranarum (B.Ranarum) is a fungus that causes an extremely rare condition known as gastrointestinal basidiobolomycosis (GIB). Human infection with B. Ranarum is described only in a few countries worldwide. We reported a 40-year-old female presented with persistent diarrhea and abdominal pain. A colonoscopy revealed cecal inflammation and ulcerations. Ultimately, a diagnosis of (GIB) was confirmed by a tissue biopsy obtained by colonoscopy, and antifungal therapy led to complete recovery without need for sugical intervention. This case contributes valuable insights to the limited literature on GIB and highlights the importance of prompt diagnosis and management in conditions that mimic more common diseases.*

## Keywords

Gastrointestinal Basidiobolomycosis, Chronic diarrhea, Splendore - Hoeppli phenomenon, Abdominal mass.

## Introduction

Basidiobolus ranarum is a known fungal species that causes Basidiobolomycosis. This fungus, typically found in soil and decaying plant matter, occasionally inhabits the gastrointestinal tracts of various animals. There are different modes of transmission in which this fungal illness is transmitted to humans through skin injuries, insect bites, as well as through inhalation. This fungus can spread to other parts of the body like the respiratory system which is the most affected organ [1]. Extrapulmonary disease can affect the skin and gastrointestinal tract System [2]. There are multiple case reports worldwide describing such a rare fungal infection. The first case of GIB was reported by Edington in 1964 in Nigeria, and since then, subsequent cases have been published globally [3]. Most cases of GIB have been reported in the United States followed by Saudi Arabia and It can affect the pediatric and adult age group and more frequently affects the male gender [4].

We report an unusual case of GIB that presented with chronic diarrhea and was initially misdiagnosed as inflammatory bowel disease, but the diagnosis of GIB was established uniquely in our case report by colonoscopy-guided biopsy. The diagnosis was reached in the previous case reports by histopathological examination after surgical resection.

## Case Presentation

A 40-year-old Saudi female was referred to our healthcare facility at King Fahad Medical City with a history of persistent diarrhea, abdominal pain, night sweats, and easy fatigability

over the last three months. Her medical history is remarkable for smoking and abdominal liposuction surgery a year ago. The patient had received a course of empirical antibiotics for chronic diarrhea without response. During the Physical examination, the patient BMI is 21. Her vitals, including her blood pressure, were measured as 95/80. Her heart rate is 80, respiratory rate is 16, and temperature is 36.8 Abdominal exam revealed a moderate right upper quadrant tenderness with palpable mass. Laboratory results showed high c-reactive protein 21 (normal range upto 5 mg/L) and erythrocyte sedimentation rate 44 (normal range for age < 20 mm/h) with normal white blood cell and including eosinophils. A contrast-enhanced CT scan revealed severe thickening in the cecum, ascending colon, and proximal transverse colon, likely representing infectious vs inflammatory conditions, Figure 1a,b. A colonoscopy showed severe congestion, inflammation, and ulcers involving the cecum while, the rest of the colon appeared normal. Also, the terminal ileum appeared normal, but there was a stricture at the ileocecal valve, Figure 2a-f. Colonosopy-guided biopsy revealed eosinophilic colitis and fungal micro-organisms consistent with B.ranarum, leading to a final diagnosis of GIB, Figure 3a-d.

A multidisciplinary meeting was conducted, where the final decision to treat with oral antifungal therapy, Itraconazole 200 mg once daily for 9 months. CT abdomen with contrast was done after 4 months of starting treatment and showed complete resolution of inflammatory mass. Also colonoscopy was reperated after 9 months with complete resolution of previous findings.

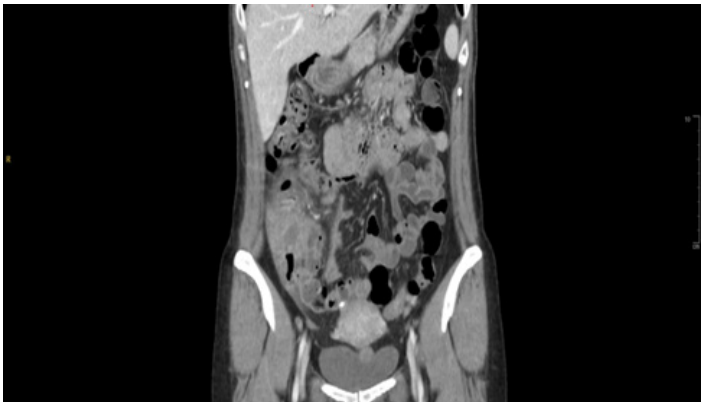

**Figure 1**: (a) A contrast-enhanced CT scans of the abdomen revealed severe thickening in the cecum, ascending colon, and proximal transverse colon.

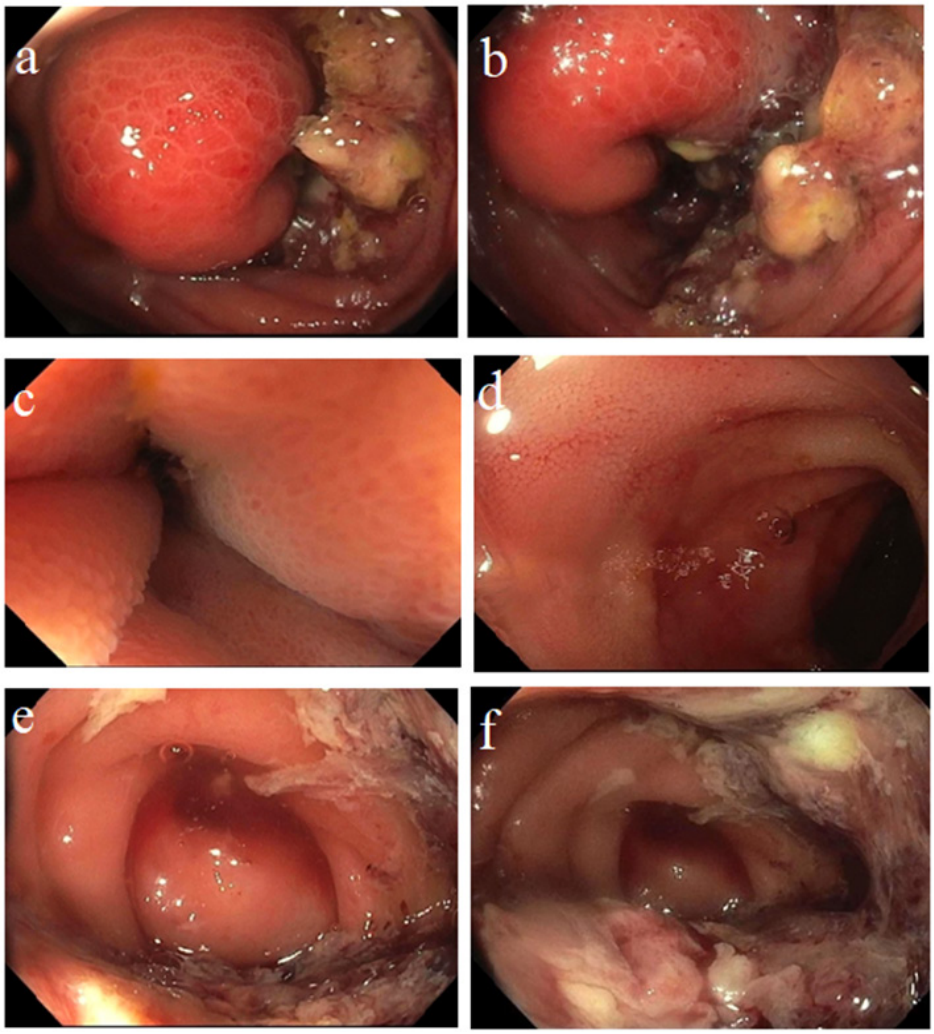


**Figure 2: (a,b):** Cecal mass like lesion with Congested ulcered mucosa. **(c, d):** Terminal Ileum appears normal. **(e,f):** Stricture at the ileocecal valve.

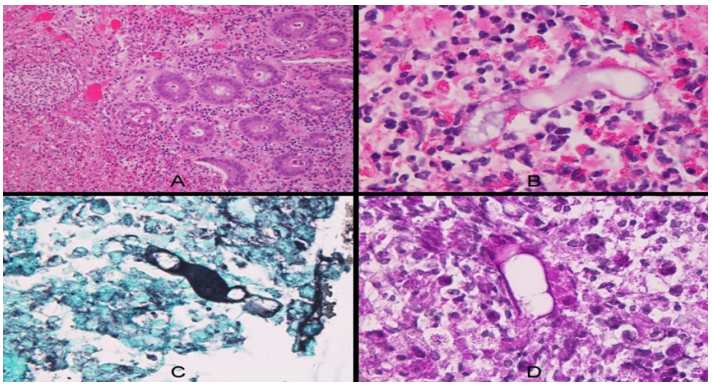


**Figure 3** (a): Dense infiltration of the colonic crypts and lamina propria by eosinophils (H&E stain, 100x magnification). (b): Fungal hyphae that are broad and irregular with hyposeptation and right-angle branching, splendore–hoeppli phenomenon (H&E stain, 400x magnification). (c): Fungal microorganisms are positive with Grocott-Methenamine silver stain (400x magnification). (d): Fungal microorganisms are positive with Periodic Acid Schiff stain (400x magnification).

## Discussion

B.Ranarum is a fungus classified within the family Entomophthoraceae of the class Zygomycetes. It is a well-documented species known for its pathological effects on skin and subcutaneous tissue. However, the involvement of extra-cutaneous sites like GIT is infrequent [5]. The majority of reported cases that have been documented have come from Middle East including Saudi Arabia, Iran, Iraq and Kuwait as well as United State [6]. The involvement of the GIT may suggest that the infection is acquired through the fecal-oral route or by ingestion of contaminated food from infected soil, even though the way of fungal entry into the host is unknown [7].

The risk factors for the fungal infection are unknown, however, earlier studies showed that the infection can be acquired following surgical procedures especially intra-abdominal procedures like inguinal hernia repair and appendectomy. Another potential risk factor is smoking. Most GIB cases have been reported in immunocompetent hosts. This disease was significantly more common in pediatrics and adolescents [8].

In a recent literature review of 102 cases of GIB, abdominal pain was the most common symptom, followed by weight loss, abdominal mass, vomiting, diarrhea, night sweats, and fever [9]. The most affected part of the GIT system is the right colon which frequently manifests as an abdominal mass that resembles inflammatory bowel disease and colon cancer [10,11]. Neutrophilia, eosinophilia, and a high ESR are frequent laboratory findings in GIB patients. In some instances, GIB can be diagnosed by stool culture and PCR analysis. However, the definitive confirmation of GIB typically relies on tissue biopsy due to its high specificity and accuracy. The immunodiagnostic test such as immunodiffusion is not standardized as it shows high specificity but low sensitivity [12]. Radiologic findings by ultrasonography and CT scan were reported as mass lesions, fistulas, and obstruction mimicking neoplastic and IBD [13,14].

The histopathologic findings that were described are granulation tissue with mixed inflammatory cells including giant cells and broad, non septated, hyphae-like structures surrounded by an eosinophilic sheath, called 'Splendore–Hoeppli phenomenon' [15]. The final diagnosis of GIB is often delayed in the majority of reported cases due to nature and rarity of the disease, as the diagnosis of IBD and colorectal cancer are the main differentials in such clinical presentation [16]. It is established histopathologically after surgical resection in the majority of cases [17,18], however in this case report, the diagnosis was proven by colonoscopy-guided biopsy without the need for surgical intervention. The standard of Care in Managment of GIB is combined antifungal therapy and surgical resection. However, the latest reports showed that the medical treatment is effective as combination therapy [18]. Here, we discuss a 40-year-old female who was diagnosed with GIB, which was initially presumed to have IBD or malignancy. Smoking and abdominal surgery were potential risk factors for GIB. In our case, colonoscopy showed a normal appearance of the ileum, which is usually involved in Crohn's disease, which was a helpful differentiating factor. 200 mg of oral Itraconazole therapy was initiated for 9 months followed by complete recovery. We believe that this case report will add valuable insights to the existing literature on gastrointestinal basidiobolomycosis and help healthcare professionals with prompt diagnosis and management of such complex conditions, mimicking other common diseases.

## Conclusion

GIB is an emerging infection, and diagnosis requires a high index of suspicion. This case serves as a reminder of the significance of considering rare fungal infections like GIB into consideration when evaluating the cause of chronic gastrointestinal symptoms, particularly in patients who have abdominal masses with eosinophilia or instances that mirror IBD but do not meet the diagnostic criteria to improve outcomes and avoid morbidity. A superior, less risky approach that may assist avoid surgical intervention in suspected cases is an endoscopic diagnosis, which involves acquiring specimens for fungal culture and histopathologic examination. Antifungal therapy can lead to complete recovery without the need to surgical resection. However, a longer duration of antifungal therapy is needed.

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