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Case Report

Gastrointestinal basidiobolomycosis masquerading as cancer☆,☆☆

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ABSTRACT

Gastrointestinal basidiobolomycosis is an unusual fungal infection caused by *Basidiobolus ranarum*, a saprophytic fungus primarily found in soil and decaying vegetables. Basidiobolomycosis typically presents as a chronic subcutaneous swelling and rarely infects the gastrointestinal tract. Thus, the infrequency of gastrointestinal infections, along with nonspecific clinical symptoms, often results in misdiagnosed cases and delays in treatment. In this article, we report the case of a 68-year-old male with gastrointestinal basidiobolomycosis masquerading as metastatic cancer. We focus on the use of radiological imaging modalities and histopathological analysis to optimize the diagnosis and treatment of this rare gastrointestinal infection.

Introduction

Gastrointestinal basidiobolomycosis (GIB) is characterized by its histopathologic appearance from surgical specimens or tissue biopsies [1]. Basidiobolomycosis was initially described as a subcutaneous, slowly developing infection primarily impacting the trunk, limbs, and buttocks of young males living in arid climates in South America, Africa, and Asia [2]. However, gastrointestinal tract infections remain perplexing. The consumption of animal feces, soil, or food contaminated by either may be responsible, but the specific mode of transmission remains unknown [1]. Common imaging findings include an abdominal mass in the colon or liver, abscess formation, and focal bowel wall thickening, causing GIB to be often mistaken for other pathologies such as intra-abdominal malignancies, inflammatory bowel disease, and diverticulitis [3]. Consideration of GIB as a differential diagnosis when patients from arid climates present with abdominal pain, weight loss, and an inflammatory mass on abdominal computed to-

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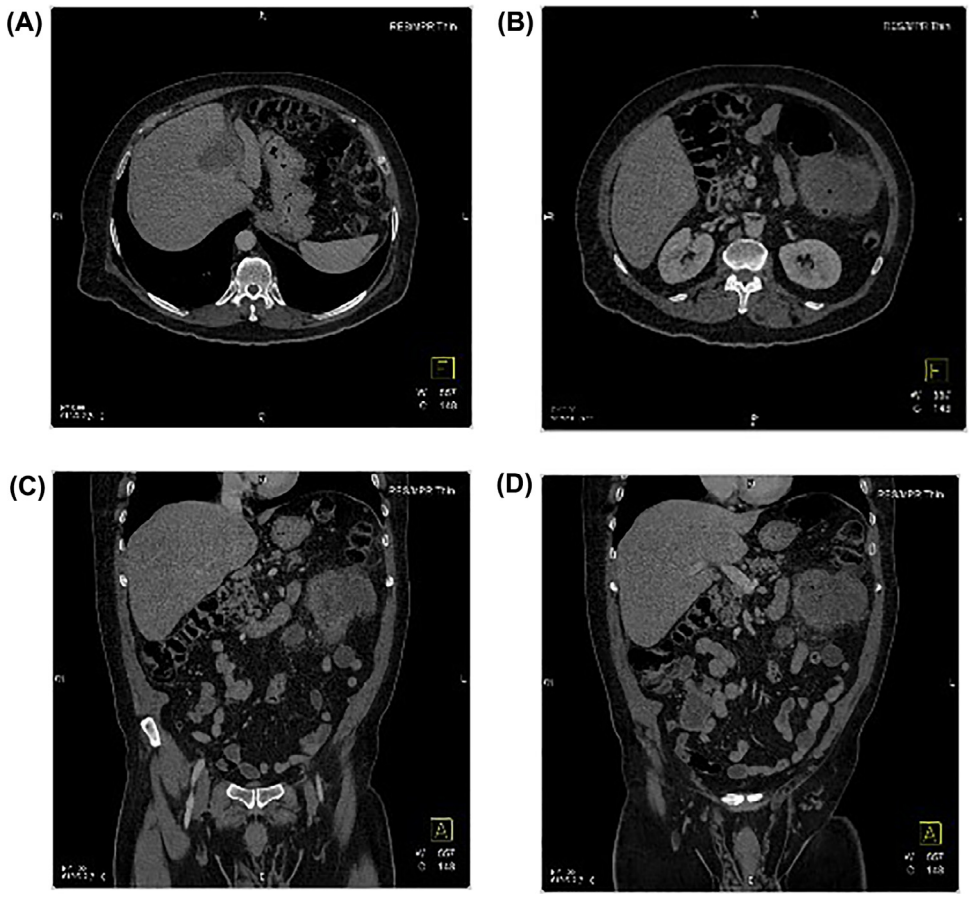

Fig. 1 – A 68-year-old male with increasing abdominal pain for the previous 10 days. Beyond abdominal distension the physical exam was unremarkable. (A) CT scan through the upper abdomen defines a 3.2 cm cystic hypodense lesion in the liver. (B) A CT scan shows evidence of a 7.7 cm necrotic process near the region of the descending colon. (C and D) Coronal CT scans defined a necrotic mass near the descending colon consistent with an inflammatory process or tumor.

mography (CT) will allow for prompt diagnosis and initiation of antifungal treatment [3]. Despite sparse data on treatment options for GIB, a literature review reveals that azole antifungal medications such as itraconazole or voriconazole, either as monotherapy or in combination with surgical resection, are effective [2,4,5].

Case presentation

A 68-year-old male with a history of diabetes mellitus type 2 presented to the ED with abdominal pain beginning 10 days prior. He described it as a constant dull ache in the left lower quadrant without aggravating or relieving factors and denied similar past episodes of pain. Upon physical examination, he appeared obese with a BMI over 30 kg/m^2, blood pressure of 142/66, and abdominal distension. The physical exam was otherwise unremarkable. A CT scan of the patient's abdomen and pelvis with IV contrast (Fig. 1) demonstrated a redundant sigmoid colon with an inflammatory mass-like area in the left mid abdomen and upper quadrant measuring up to 9.6 × 7.7 × 7.4 cm with downstream obstructive changes. Further, visible within the adjacent mesentery was a 3.2 cm rounded lesion with characteristics similar to the colonic focus representing a metastatic deposit or phlegmon associated with diverticulitis. A dominant hypoenhancing lesion in the hepatic dome extended down towards the porta hepatis, measuring 5.0 × 3.6 × 3.2 cm, and a subtle enhancing 1.4 cm nodule was observed in the right hepatic lobe. These hypoattenuating hepatic lesions reflected "abscesses or metastases." Given the appearance on the CT and the patient's history of a clear colonoscopy within 3 years, the findings suggested smoldering diverticulitis, supported by the patient's leukocytosis. He was empirically treated for diverticulitis with IV ceftriaxone/metronidazole without improvement.

A repeat CT scan of the abdomen and pelvis with IV contrast (Fig. 2) showed the mass infiltrating the colon wall. The liver lesion appeared to be necrotic (extending to the porta hepatis measuring 4.6 × 6.1 cm, previously 3.8 × 5.2 cm). Mild ductal dilatation was seen within the adjacent left lobe and a subtle lesion within the posterior right hepatic lobe measured 2.2 cm (slightly increased). Further, the pancreas had atrophied, and a punctate left renal cortical cyst was present. A mass involving the proximal sigmoid colon with irregular nodular wall thickening and adjacent stranding appeared

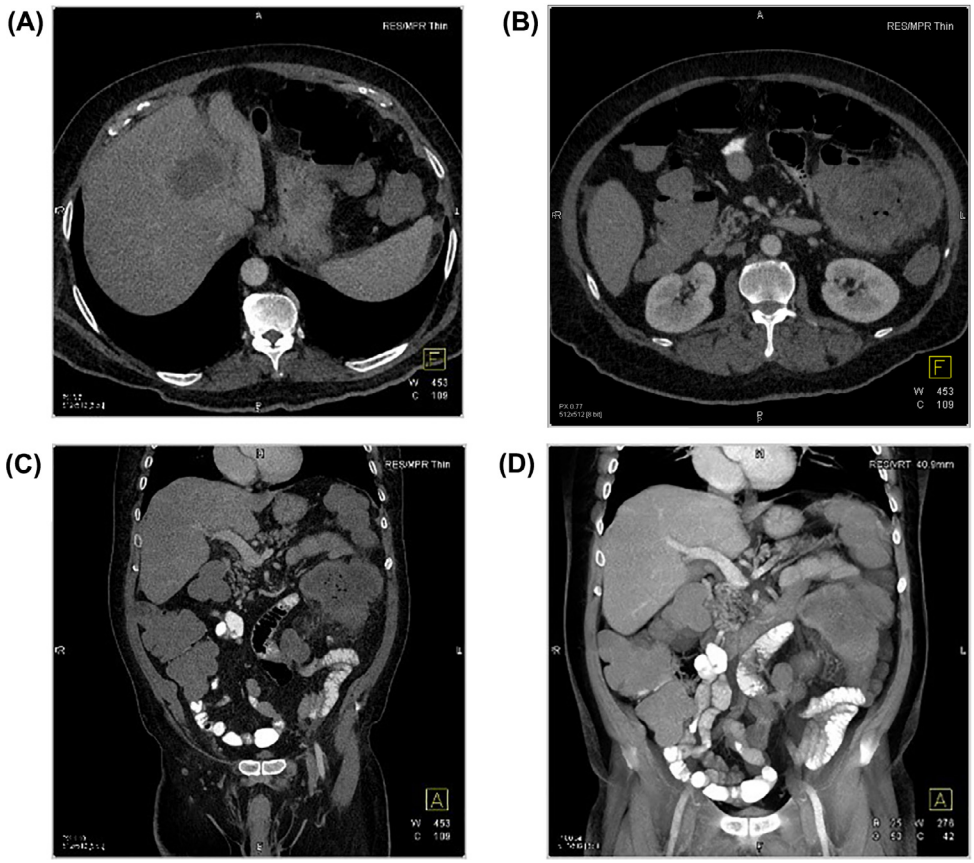

Fig. 2 – Follow-up CT post treatment without improvement. (A) The liver mass or abscess has increased in size since the prior exam. (B) The necrotic mass involving the bowel has also expanded in size and inflammation has also increased since the prior study. (C and D) Coronal and 3D VRT images define the necrotic mass with air bubbles and inflammation. Differential diagnosis still included inflammatory vs. neoplastic processes. The colon was not obstructed.

again. The mass measured 7.4 × 11.3 cm in transaxial dimension, previously 7.4 × 10.0 cm. The necrotic mass/adenopathy within the adjacent mesentery measured 2.9 × 3.9 cm (slightly increased). Also noted was a small level of pelvic ascites, which had increased. The hepatic lesions were concerning for metastatic colorectal cancer. A flexible sigmoidoscopy showed the lesion to be near obstructing; therefore, the patient was likely to undergo a surgical resection. An abdominal X-ray displayed persistent colonic dilation. Repeated blood cultures were negative, but complete blood counts indicated that the patient had eosinophilia.

To assess whether the lesions were metastatic, a flexible sigmoidoscopy with biopsies and an ultrasound-guided core liver biopsy were performed; both were negative for malignancy. The patient then underwent an IR-guided biopsy of the mesenteric mass next to the sigmoid colon, with pathological findings negative for malignancy and suggestive of an infectious etiology. The biopsy supported an infectious etiology and demonstrated acute as well as chronic inflammation, including eosinophils, neutrophils, giant cells, and granuloma inflammation. GMS and PAS stains highlighted fungal organisms and the tissue showed abundant eosinophils and the Splendore-Hoeppli phenomenon, which are common morphological features in *Basidiobolus ranarum*. Notably, basidiobolomycosis can also present with intestinal tumor-like masses and other intestinal symptoms. Based on morphologic features, intestinal Mucorales genera (mucormycosis) were also in the differential diagnoses, although the histologic picture was not classic.

In a follow-up consultation, the patient noted that over the preceding 5 months, he had difficulty with his bowels and measured a 20-30 pound weight loss over the previous 6-8 months. He also revealed having spent a month in Iran, an area with endemic gastrointestinal basidiobolomycosis. He denied any raw meat ingestion, other sick contacts, or other viral exposures. Subsequently, the patient was started on itraconazole with no indication for urgent surgical intervention. Nonresolution with medical management would leave surgical clearance as an option, though. The patient has had 2 subsequent abdominal X-rays, neither of which revealed evidence of obstruction.

Discussion

GIB is an exceptionally rare presentation of the basidiobolus sp. infection. According to a review of 102 GIB cases world-

wide, the most frequent clinical manifestation was abdominal pain (86.6%), followed by weight loss (33.3%), abdominal distension (16.7%), vomiting (15.7%), and then diarrhea (13.7%), with 40.2% of patients reporting with a fever [1]. Our patient presented with the 3 most common aforementioned symptoms, though he did not develop a fever until 3 weeks after admission. The nonspecificity of these symptoms and common occurrence in immunocompetent patients render GIB challenging to diagnose, which often results in delays in treatment. Patients frequently have neutrophilic leukocytosis and eosinophilia, which was consistent with our case as leukocytosis persisted irrespective of antibiotics [6]. As of 2018, approximately 122 basidiobolomycosis cases have been reported worldwide, with most from Middle Eastern countries (ie, Iran, Saudi Arabia, and Kuwait) [7,8]. Available data indicates males may be more susceptible than females [9], but there is no specific age group associated with GIB, as reported ages range from 1.5 years to 80 years [2].

A review of GIB cases concluded that a mass in abdominal organs was the most common finding on CT scans and endoscopy [2]. On abdominal and pelvic CTs, it was reported that a colorectal mass, as seen in our patient, was the most frequent finding (48%), followed by a liver mass (20%), and a mass on the small intestine (11%) [3]. Typically, GIB affects the colon, liver, and small bowel but can also impact the pancreas and biliary tract [10–14]. A review of the epidemiology, histopathology, management, clinical manifestations, and prognosis of GIB revealed that the colon and rectum (82%), small intestines (36%), as well as liver and gallbladder (30%), were the most frequent sites of involvement [2]. Our case, wherein gastrointestinal involvement included the sigmoid colon, liver, and small intestines, is consistent with the findings in the abovementioned article. These discoveries are commonly mistaken for neoplasms or other gastrointestinal conditions such as intra-abdominal malignancy (43%), inflammatory bowel disease (16%), or diverticulitis (11%) [2].

Currently, there are no specific treatment guidelines for GIB, but some authors have noted that antifungal medications alone or in combination with surgical resection of masses can be effective [2,4,5]. For our patient, surgical resection remained an option as he underwent antifungal treatment. Although surgery can be effective, there is uncertainty about the exact role that it plays in the treatment of GIB patients. Therefore, medical therapy before surgical intervention is recommended [2,15].

Conclusion

GIB should be considered as a differential diagnosis in patients with abdominal pain, weight loss, eosinophilia, and an inflammatory mass unresponsive to nonantifungal therapies, especially if the patient comes from Iran, Saudi Arabia, Kuwait, or arid regions of the United States (mainly Arizona) [2]. Diagnosis of GIB is typically by fungal staining of biopsy tissue or culture (negative in our case). Histopathologically, characteristic features are the presence of necrotizing granulomatous inflammation, tissue eosinophilia, and bright, intensely eosinophilic granular material (Splendore-Hoeppli phenomenon) [2]. Although the Splendore-Hoeppli phenomenon can be seen in various infectious conditions, GIB diagnosis should be considered when these histological characteristics and previously mentioned clinical symptoms are seen in male patients from endemic areas [16].

Author contribution

All authors contributed equally to the writing of this manuscript.

Patient consent

The patient reported in the manuscript signed the informed consent/authorization for participation in research, which includes the permission to use data collected in future research projects such as the presented case details and images used in this manuscript.

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