OPEN

Gastrointestinal Basidiobolomycosis in a Child: Unusual Fungal Infection Mimicking Eosinophilic Gastrointestinal Diseases—A Case Report and Review of the Literature

*Fat'hiya Al Harthy, MD, †Omaima Al Kalbani, MD, *Mohammed Al Masqri, MD, †Ahmed Al Dhuhli, MD, and †Khoula Al Said, MD

Abstract: Gastrointestinal basidiobolomycosis (GIB) is unusual, rare, and emerging fungal infection. It is caused by *Basidiobolus ranarum*. Unlike other fungal infections, *B ranarum* affects immunocompetent individuals with potentially grave sequelae if unrecognized. GIB is difficult to be diagnosed due to unspecific clinical presentation. Diagnosis of basidiobolomycosis requires culture of *B ranarum* from tissue. Optimal management of GIB includes antifungal therapy alongside if necessary early surgery.

Key Words: gastrointestinal basidiobolomycosis, fungal infection, Oman

INTRODUCTION

Herein, we report a pediatric case of gastrointestinal basidiobolomycosis (GIB) in previously healthy Omani male with a unique presentation that mimics eosinophilic gastrointestinal diseases. The diagnosis was established by a histopathology tissue, treated successfully with antifungal therapy and surgical resection of the affected bowel segments.

CASE PRESENTATION

Herein, we report an 8-year-old Omani male previously healthy presented with 2 weeks history of bloody diarrhea, right iliac fossa (RIF) abdominal pain, low grade fever, and nonbilious vomiting. He had a longstanding history of intermittent abdominal pain and significant weight loss ~6 kg in 2 months period. He had also reported a history of dysphagia to solid food over the same period. He had otherwise reported no extra gastrointestinal manifestations. There was no history of a recent travel or contact with a patient with tuberculosis (TB) or coronavirus disease 2019. There was a family history of allergy but no family history of inflammatory bowel disease, autoimmune disorders, TB, or malignancies.

On physical examination, he was pale, severally malnourished with no lymphadenopathy. He had a mild tenderness on abdominal palpation at RIF. Examination of other systems showed no abnormality.

Laboratory investigations as shown (Supplemental Digital Content Table 1, http://links.lww.com/PG9/A57) are microcytic hypochromic anemia with normal white blood cell counts but peripheral eosinophils and thrombocytosis, high inflammatory marker, low albumin level, and elevated IgE level. He had a positive occult blood and fecal caloprotectin was 218 mg/kg. Stool infectious samples showed only positive for *Clostridium difficile*. His HIV, TB, and coronavirus disease 2019 screening tests were negative. Immunological, autoimmune, and tumor markers were unremarkable.

He was treated successfully for *C difficile* infection. As he had dysphagia, nasogastric tube feeding was initiated during hospitalization.

The radiological investigations included abdominal ultrasounds and computed tomography scan of the abdomen which showed marked circumferential wall thickening affecting the distal ileal loops, ileocecal valve, cecum, and right-sided colon up to the splenic flexure of the colon (the thickness was around 1–1.6 cm; Fig. 1). Multiple enlarged regional mesenteric lymph nodes, edema, mild intraperitoneal fluid, and inflammatory fat stranding were noted. There was no abscess collection, fistula formation, or masses seen.

Esophagogastroduodenoscopy was unremarkable with no evidence of eosinophilic esophagitis (Fig. 2A). The colonoscopy revealed normal colonic and terminal ileum mucosa (Fig. 2B, C).

Histopathologic examination of biopsies demonstrated numerous eosinophils (>20/high power field) in all biopsies (Fig. 3A, B). There were no microorganisms identified.

Patient was initially treated with a possible diagnosis of eosinophilic gastrointestinal diseases or a possibility of an initial presentation of Crohn disease. Therefore, he was started on IV methylprednisolone.

A week after starting IV methylprednisolone, his overall clinical and biochemical condition improved. However, a week later, his abdominal pain reoccurred with a palpable, mobile, and tender mass at RIF. Hence, a repeated ultrasound showed interval progression in the degree of the bowel wall thickening with interval progression in the surrounding inflammatory changes with pericolonic fluid collection in which perforation was suspected at that time. Therefore, IV steroid discontinued, and he underwent an exploratory laparotomy which identified a large mass involving terminal ileum, right colon, and extending to the hepatic flexure. Resection of ileocecal valve and right hemicolectomy done with ileotransverse anastomosis.

Histopathology examination of the mass revealed fungal organisms with broad, pleomorphic hyphae, and occasional septa surrounded by an eosinophilic material (Splendore-Hoeppli phenomenon; Fig. 3C). Dense eosinophilic infiltrate with scattered foreign body giant cells is identified around the organisms. The fungi were seen in all the colonic layers except the mucosa.

Received May 17, 2021;accepted July 2, 2021.

*Division of Gastroenterology & Nutrition, Department of Pediatric, Royal Hospital, Muscat, Sultanate of Oman; and †Ministry of Health, Muscat, Sultanate of Oman.

The authors report no conflict of interest.

Parental knowledge and verbal consent was obtained before manuscript submission and consent given for the case report publication.

Supplemental digital content is available for this article.

Correspondence: Fat'hiya Al Harthy, MD, Division of Gastroenterology & Nutrition, Department of Pediatric, Royal Hospital, Ghala St, Muscat, Sultanate of Oman. E-mail: fathiya.al-harthy@medportal.ca

JPGN Reports (2021) 2:3(e109)
ISSN: 2691-171X
DOI: 10.1097/PG9.0000000000000109

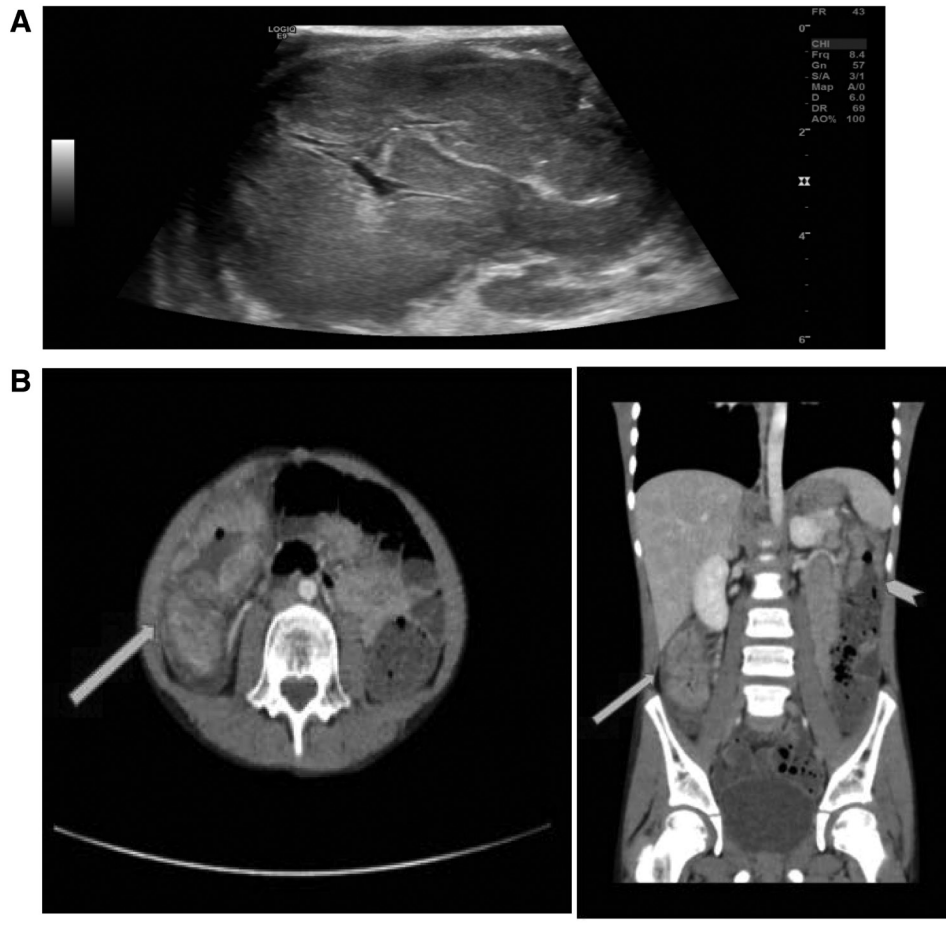

FIGURE 1. Radiological images. A) Ultrasound of the abdomen showed circumferential wall thickening of the cecum with severely narrowed lumen. B) CT scan of the abdomen with contrast (axial and coronal images) showed marked wall thickening of cecum (arrow) up to the splenic flexure (small arrow).

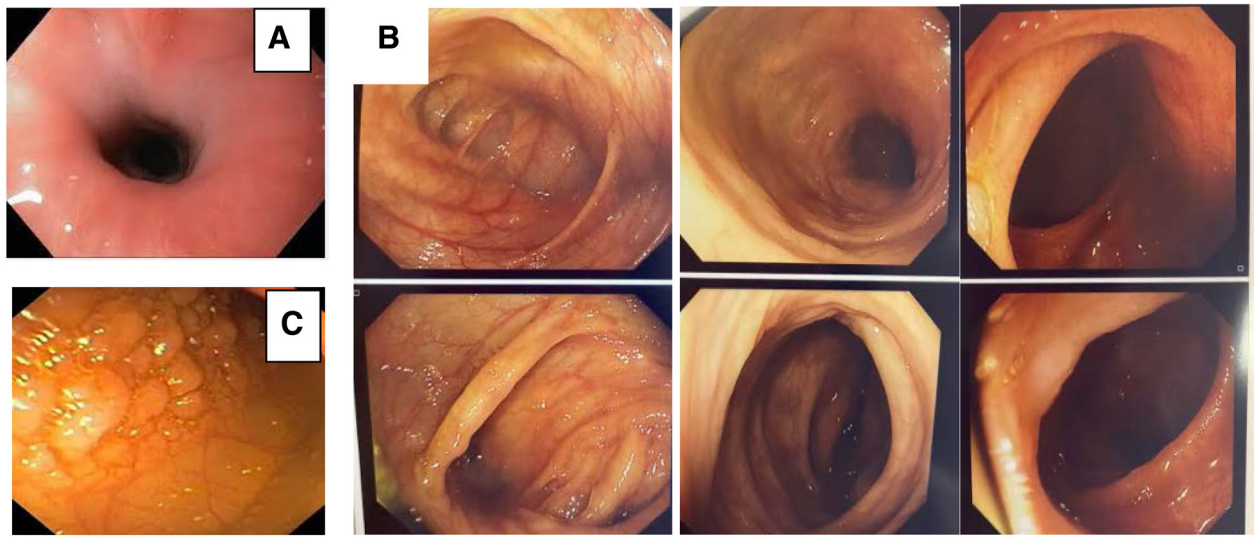

FIGURE 2. EGD and colonoscopy. A) Normal esophagus, (B) normal colon, (C) normal terminal ileum. EGD = esophagogastroduodenoscopy.

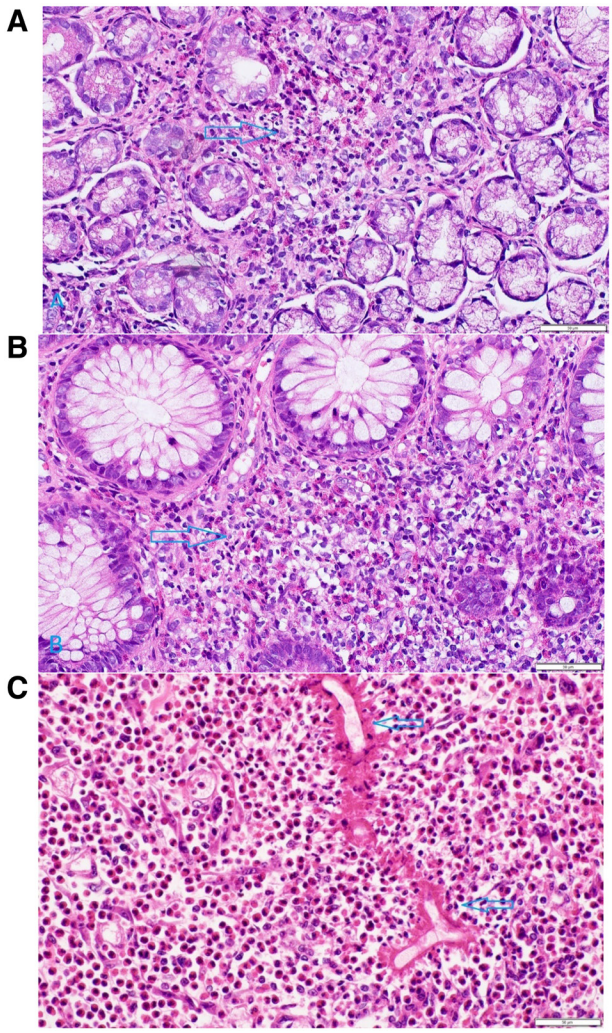

FIGURE 3. Intermediate power view (×20) showing gastric (A) and colonic (B) mucosa with numerous eosinophils in the lamina propria. Intermediate power view (×20) showing the typical morphology of Basidiobolus fungi (C).

Based on the histopathologic tissue findings, a diagnosis of GIB was established. Subsequently, antifungal treatment was initiated with IV voriconazole a dose of 8 mg/kg/dose twice daily for a total of 8 months.

On clinical visit follow-ups, he had regained his weight and was asymptomatic. His biochemical markers have normalized (Supplemental Digital Content Table 1 http://links.lww.com/PG9/A57).

DISCUSSION

GIB is a newly identified disease with 122 cases reported worldwide as of 2018 of which 46 cases reported in adults and most pediatric cases came from Saudi Arabia (37.2%) followed by United States (21.6%) and Iran (20.6%) (1).

GIB disease rarity combined with its broad symptoms and potential for clinical mimicry of more common clinical entities make its early recognition exceedingly challenging (2). Yet, it is unclear how *B ranarum* gains access to the host's gastrointestinal tract. It is hypothesized that ingestion of food contaminated by the fungus from soil or animal excreta is the most likely route of infection in GIB. It typically affects immunocompetent individuals with potentially grave sequelae if left untreated (3).

The most common presenting symptom of GIB is abdominal pain (86.3%), followed by fever (40.2%), weight loss (33.3%), abdominal mass (30.4%), vomiting (15.7%), and then diarrhea (13.7%) (1). In addition, this study review concluded that peripheral blood eosinophilia was detected in 85.7% of cases (1).

Diagnosis of GIB is challenging owing to its nonspecific clinical presentation and rarity. A recent review of 102 cases of GIB was misdiagnosed as neoplasm and inflammatory bowel disease in two-thirds of patients (4). A definitive diagnosis of basidiobolomycosis requires a microbial culture of *B ranarum* from fresh aspiration or histopathological examination (4). Microbiology cultures remain the gold standard of diagnosis, although this is achieved in only 50% of cases (5). Histopathological examinations usually exhibit broad, thin-walled, hyposeptated hyphae surrounded by a dense eosinophilic material (Splendore-Hoeppli phenomenon) which is a characteristic for basidiobolomycosis (4,5). The use of polymerase chain reaction and ELISA has been recently reported in the diagnosis of GIB (5). Polymerase chain reaction showed high sensitivity and specificity but still not widely used due to the rarity of the disease.

A favorable outcome of GIB depends on early diagnosis, antifungal therapy, and surgical debulking. Mortality from GIB is high with rates reaching 16% if untreated (1). Misdiagnosis and delay to initiate appropriate treatment may lead to disseminated disease that can result in an overly complicated course and reduced response to antifungal treatment. The most frequent antifungal used in treating GIB is itraconazole (73%), followed by amphotericin (22%), ketoconazole (8%), and voriconazole (5%) (6). Voriconazole is the safest and tolerable drug among the rest. In the other hand, amphotericin has been linked with several clinical failures. Surgical resection of the inflammatory mass combined with antifungal therapy is considered standard treatment of GIB, though successful outcome with antifungal treatment alone is well described (7). Corticosteroid may not be a good choice for the treatment of GIB, although often it was started in most of the reported cases as these cases present with a challenging diagnostic dilemma.

In conclusion, we presented a case of GIB. The challenge in reaching GIB diagnosis highlights the importance of increasing awareness and early recognition of this condition. Without a high index of clinical suspicion and clinician familiarity, GIB will continue to be a missed diagnosis with potentially grave consequences.

ACKNOWLEDGMENTS

The authors acknowledge Dr Ibtisam Al Shuiali who contributed to the editing and obtaining radiological images for the manuscript.

REFERENCES

1. Pezzani MD, Di Cristo V, Parravicini C, et al. Gastrointestinal basidiobolomycosis: an emerging mycosis difficult to diagnose but curable. Case report and review of the literature. *Travel Med Infect Dis.* 2019;31:101378.
2. Shreef K, Saleem M, Saeedd MA, et al. Gastrointestinal basidiobolomycosis: an emerging, and a confusing, disease in children (a multicenter experience). *Eur J Pediatr Surg.* 2018;28:194–199.
3. Kurteva E, Bamford A, Cross K, et al. Colonic basidiobolomycosis—an unusual presentation of eosinophilic intestinal inflammation. *Front Pediatr.* 2020;8:142.
4. Balkhair A, Al Wahaibi A, Al-Qadhi H, et al. Gastrointestinal basidiobolomycosis: beware of the great masquerade a case report. *IDCases.* 2019;18:e00614.
5. Omar Takrouni A, Heitham Schammut M, Al-Otaibi M, et al. Disseminated intestinal basidiobolomycosis with mycotic aneurysm mimicking obstructing colon cancer. *BMJ Case Rep.* 2019;12:e225054.
6. Almoosa Z, Alsuhaibani M, AlDandan S, et al. Pediatric gastrointestinal basidiobolomycosis mimicking malignancy. *Med Mycol Case Rep.* 2017;18:31–33.
7. Ahmed Albishri A, Shoukeer MAH, Shreef K, et al. Gastrointestinal basidiobolomycosis. *J Pediatr Surg Case Rep.* 2020;55:1014114.