RESEARCH ARTICLE

Disseminated gastrointestinal basidiobolomycosis: Case report

Mona Al-Shahrani[1], Awad Mohammed Al-Qahtani[2]

[1]Department of Internal Medicine, King Khalid University, Abha, Saudi Arabia, [2]Department of Family & Community Medicine, Faculty of Medicine, Najran University, Najran, Saudi Arabia

Abstract

Gastrointestinal basidiobolomycosis (GIB) is an invasive fungal infection affecting immunocompetent children and young adults. Most of the reported cases are from the southwestern region of Saudi Arabia; it has a non-specific presentation which can mimic inflammatory bowel disease, gastrointestinal tuberculosis, and colon cancer. GIB usually affects the colon and small intestine while disseminated GIB is very rare. Liver involvement was reported in only 5 cases. We report here the case of a 22-year-old Saudi girl living in Jizan (southern region of Saudi Arabia) presented with disseminated gastrointestinal intramural and liver abscesses which were successfully treated with the aspiration of liver abscess and oral antifungal therapy (itraconazole). GIB diagnosis need a high index of suspicion and should be considered in patients from the southwestern region of Saudi Arabia with gastrointestinal intramural and liver abscesses. GIB is an emerging infection that might lead to diagnostic confusion, morbidity, and mortality.

Key words: Fungal infection, Gastrointestinal basidiobolomycosis, Itraconazole, Liver abscess

INTRODUCTION

Basidiobolomycosis is an unusual fungal infection that manifests in the skin and soft tissue and rarely involves other systems.[1] Basidiobolomycosis is a common infection that develops following traumatic inoculation of the fungus under the skin with high prevalence in the tropical and subtropical regions of the world. The most favored hypothesis for transmission of this gastrointestinal infection is through consumption of contaminated food or dirt, although the exact mode of the acquisition remains poorly understood.[2,3] 19 cases of GIB were reported in the U.S. since 1986, out of these 17 cases were from Arizona and one case from a southern Utah town bordering Arizona. 11 cases were also reported from Saudi Arabia, indicating that these rare infections are more common in arid and desert regions.[4] The present case study, reported in May 2015, describes a rare case of GIB in a Saudi patient who was treated successfully.

CASE REPORT

A 22-year-old Saudi girl from Sapia (south region of Saudi Arabia), known to have type 2 diabetes mellitus presented to Aseer Central Hospital with a 4-month history of diarrhea more than 4 times a day, associated generalized abdominal pain and distention. She admitted history of fever on and off and unintentional significant weight loss around 10 kg with loss of appetite. There was no jaundice or rectal bleeding. On examination, she was underweight 35 kg, afebrile, blood pressure: 95/60 mm Hg, pulse: 110, 95% O_2 on room air (RA). Pale, no lymphadenopathy, and no jaundice. Abdomen was largely distended, tympanic resonant in percussion, no organs or masses could be felt.

The laboratory report revealed high erythrocyte sedimentation rate (125 mm/h), high alkaline phosphatase (168 IU/L) and gamma-glutamyl transpeptidase (63 IU/L), and albumin level (1.9 g/dL). A computed tomography (CT) scan of abdomen revealed marked wall thickening of transverse colon, and the upper two-third of the descending

Address for correspondence:
Dr. Awad Mohammed Al-Qahtani,
Department of Family & Community Medicine, Faculty of Medicine, Najran University, Najran, Saudi Arabia
Tel: 00966530540450, Fax: 0175417111.
E-mail: awadresearch17@gmail.com

Received: 28-11-2017
Revised: 14-12-2017
Accepted: 20-12-2017

colon is noted, consequent marked luminal narrowing has been identified, resulting in marked dilatation of the ascending and proximal part of the transverse colon. There is marked thickening of the terminal ileal loops. In addition, right hepatic lobe mass lesion was seen of the complex cystic nature adjacent to gallbladder [Figure 1]. It measures 78 mm × 33 mm in cross-section. All these findings are suggestive of an infectious process involving the colon and right hepatic abscess.

Circumferential marked wall thickening in the transverse colon and upper two-third of the descending colon. Consequent marked luminal thinking has been identified.

This is resulting in marked dilatation of the ascending colon and proximal part of the transverse colon. In addition, right hepatic lobe mass is seen near to the gallbladder, it measured 78 mm × 33 mm in cross-section.

Upper and lower gastrointestinal endoscopy reveals submucosal swelling in the gastric body (greater curvature) about 4 cm × 4 cm with normal mucosa above it, and discharging pus with biopsy. There are also cystic lesions all over the ascending, transverse, and descending colon. These cysts are discharging pus [Figure 2].

A biopsy was taken from terminal ileum, and histopathology report showed heavy infiltration of the lamina propria by chronic inflammatory cells and eosinophils, with granuloma formation of multinucleated giant cells. Thick fungal hyphae were detected consistent with *Basidiobolus ranarum*. A percutaneous liver biopsy was done under ultrasound guidance which revealed budding yeast cells [Figure 3].

Treatment was initiated immediately with itraconazole at a dose of 100 mg twice daily. 2 weeks after initiation of itraconazole therapy, diarrhea stopped and patient appetite improved along with her general condition.

Follow-up CT Scan

CT scan revealed a decrease in the size of the liver abscess and completes resolving of the wall thickness of the colon despite persisting of the dilatation of the colon [Figure 4].

DISCUSSION

Basidiobolomycosis is a rare fungal infection caused by *B. ranarum*. It usually manifests with subcutaneous infection, gastrointestinal lesions and rarely with systemic affection.[5] GIB is an invasive fungal infection that leads to substantial morbidity and mortality and diagnostic confusion.[6] "The main differential diagnosis of GIB with granuloma includes inflammatory bowel disease, intestinal tuberculosis, sarcoidosis, amebiasis, and malignancy."[7]

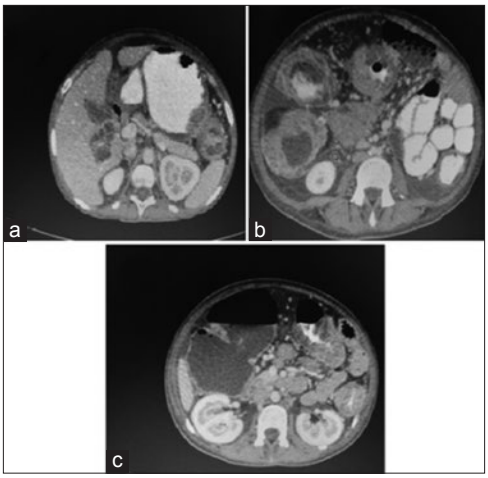

Figure 1: (a-c) Computed tomography scan of abdomen revealed marked wall thickening of transverse colon, and the upper two-third of the descending colon is noted, consequent marked luminal narrowing has been identified, resulting in marked dilatation of the ascending and proximal part of the transverse colon

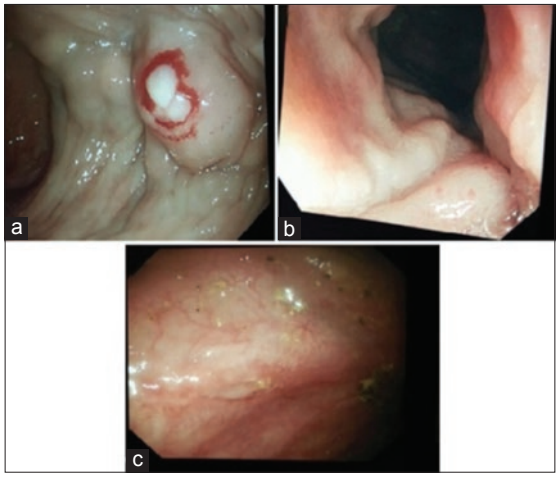

Figure 2: (a-c) Upper and lower gastrointestinal endoscopy showing submucosal swelling in the gastric body about 4 cm× 4 cm with normal mucosa above it, and discharging pus with biopsy

Characteristic histopathological features of GIB include mixed suppurative and granulomatous inflammation accompanied with prominent tissue eosinophilia. Histochemical stains reveal the fungal structures surrounded by homogeneous eosinophilic material and histiocytes, i.e., the Splendore–Hoeppli phenomenon.[8] Supporting the supposition of a gastrointestinal portal of entry is the frequent finding on the pathology of intestinal thickening with transmural inflammation.[9] On endoscopy, cobblestoning of the intestinal mucosa has been described, similar to Crohn's disease,[10] and conversely, the superficial mucosa has been described as intact in several cases. This may be analogous to the finding of epidermal preservation in cases of subcutaneous basidiobolomycosis,[8] suggesting that the organism gains entry through a mucosal surface, but spreads through submucosal layers. There have been scant

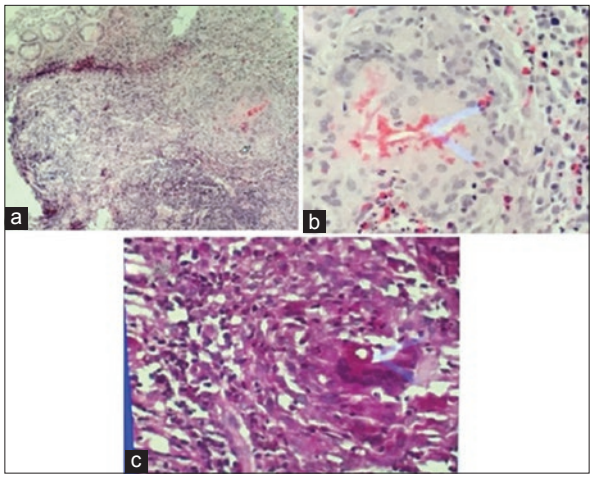

Figure 3: (a-c) Duodenal biopsy revealed heavy infiltration of the lamina propria by chronic inflammatory cells and eosinophils, with granuloma formation of multinucleated giant cells

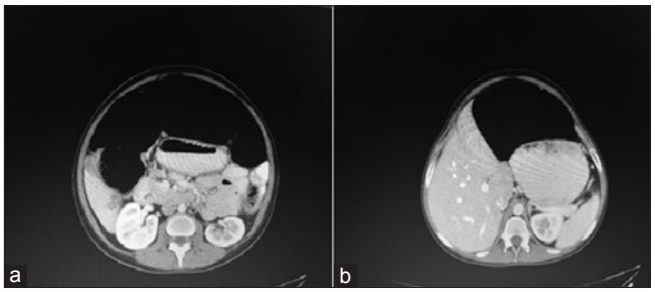

Figure 4: (a-b) Follow-up computed tomography scan revealed a decrease in the size of the liver abscess and complete resolving of the wall thickness of the colon

reports of intra-abdominal basidiobolomycosis arising from a cutaneous focus, but the majority of cases appear to start with an intramural mass or abscess with contiguous spread to surrounding organs, as occurred in the patient.[7]

Although the gastrointestinal form of basidiobolomycosis is an increasingly reported manifestation of this unusual infection, the diagnosis often proves elusive due to then non-specific clinical presentation. In a recent review, abdominal pain was the most common presenting symptom (84%), followed by an abdominal mass (43%) and constipation (39%); fever was only present in 32% of cases.[11] Based on the radiographic appearance of an abdominal (usually colonic) mass with spread to surrounding organs, including fertilization, the disease often mimics colon cancer or Crohn's disease which may delay diagnosis.[12]

In culture negative cases, serology for *B. ranarum* may have a role; based on limited studies, the sensitivity is estimated to be 50%, and titers may correlate with response to therapy. Recently, sequencing of a polymerase chain reaction (PCR) product from tissue has been increasingly utilized as an alternative means of diagnosis. In this case, the DNA for PCR was extracted from a paraffin-embedded surgical specimen, since no cultures were sent at the time of surgery due to low clinical suspicion for infection.

A standard treatment regimen for GIB has not yet been established. Many patients are treated with a combination of surgical and medical therapies. Although clinical improvement with antifungal therapy alone has been reported, the long-term prognosis in such cases remains poorly understood. Antifungal therapy includes itraconazole which is most frequently used (73%), followed by amphotericin (22%), ketoconazole (8%), and voriconazole (5%). However, amphotericin therapy has been associated with several adverse effects and clinical failures. Average duration for antifungal therapy is 8 months, and overall survival rate is estimated at 80%.[13]

CONCLUSION

The present report concludes that even though GIB is a rare disease, special attention should be given to patients with abdominal distention and persistent diarrhea coming from the southwestern region of Saudi Arabia. The early diagnosis of this condition is particularly crucial. The most-preferred treatment for GIB includes both surgical resection and appropriate antifungal therapy. The azole antifungal group remains the drug of choice for treating GIB, including ketoconazole, itraconazole, and voriconazole. Awareness of its clinical presentation, radiographic and endoscopic findings and characteristic histopathological findings can facilitate prompt diagnosis and initiation of effective antifungal therapy. Persistent eosinophilia in patients presenting with any of the above clinical findings should alert clinicians to the possibility of GIB. Culture of involved tissues and stool with appropriate fungal growth media are useful diagnostic adjuncts. Further researches about molecular detection of human fungal pathogens are urged as they can definitely settle disputed diagnosis.

REFERENCES

1. Al-Maani AS, Paul G, Jardani A, Nayar M, Al-Lawati F, Al-Baluishi S, *et al.* Gastrointestinal basidiobolomycosis: First case report from oman and literature review. Sultan Qaboos Univ Med J 2014;14:e241-4.
2. Al Asmi MM, Faqeehi HY, Alshahrani DA, Al-Hussaini AA. A case of pediatric gastrointestinal basidiobolomycosis mimicking crohn`s disease. A review of pediatric literature. Saudi Med J 2013;34:1068-72.
3. Healthcanal. Mayo Clinic Reports Emerging Fungal Infection in Southwest That Mimics Cancer. Available from: http://www.healthcanal.com/infections/28101-Mayo-Clinic-Reports-Emerging-Fungal-Infection-Southwest-That-Mimics-Cancer.html. [Last accessed on 2017 May 30].
4. Alltucker K. Mayo Study: Rare Fungal Infection Emerges in Arizona. The Arizona Republic. Available from: http://www.archive.azcentral.com/arizonarepublic/local/articles/2012/03/29/20120329mayo-study-rare-fungal-

infection-found-arizona.html. [Last accessed on 2017 Jun 06].
5. Al Jarie A, Al Azraki T, Al Mohsen I, Al Jumaah S, Almutawa A, Mohd Fahim Y, *et al.* Basidiobolomycosis: Case series. J Mycol Med 2011;21:37-45.
6. Al Jarie A, Al-Mohsen I, Al Jumaah S, Al Hazmi M, Al Zamil F, Al Zahrani M, *et al.* Pediatric gastrointestinal basidiobolomycosis. Pediatr Infect Dis J 2003;22:1007-14.
7. El-Shabrawi MH, Kamal NM, Kaerger K, Voigt K. Diagnosis of gastrointestinal basidiobolomycosis: A mini-review. Mycoses 2014;57 Suppl 3:138-43.
8. Cazorla A, Grenouillet F, Piton G, Faure É, Delabrousse É, Mathieu P, *et al.* A letal case of gastro-intestinal basidiobolomycosis. Ann Pathol 2014;34:228-32.
9. Hussein MR, Musalam AO, Assiry MH, Eid RA, El Motawa AM, Gamel AM, *et al.* Histological and ultrastructural features of gastrointestinal basidiobolomycosis. Mycol Res 2007;111:926-30.
10. AlSaleem K, Al-Mehaidib A, Banemai M, bin-Hussain I, Faqih M, Al Mehmadi A, *et al.* Gastrointestinal basidiobolomycosis: Mimicking crohns disease case report and review of the literature. Ann Saudi Med 2013;33:500-4.
11. Nemenqani D, Yaqoob N, Khoja H, Al Saif O, Amra NK, Amr SS, *et al.* Gastrointestinal basidiobolomycosis: An unusual fungal infection mimicking colon cancer. Arch Pathol Lab Med 2009;133:1938-42.
12. Saadah OI, Farouq MF, Daajani NA, Kamal JS, Ghanem AT. Gastrointestinal basidiobolomycosis in a child; an unusual fungal infection mimicking fistulising crohn's disease. J Crohns Colitis 2012;6:368-72.
13. Albaradi BA, Babiker AM, Al-Qahtani HS. Successful treatment of gastrointestinal basidiobolomycosis with voriconazole without surgical intervention. J Trop Pediatr 2014;60:476-9.

Source of Support: Nil. **Conflict of Interest:** None declared.