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CLINICAL CASE REPORT

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A Case Report of Gastrointestinal Basidiobolomycosis Treated With Voriconazole

A Rare Emerging Entity

Awaji Qasim AL-Naemi, MBBS, Liaqat Ali Khan, MBBS, Ibrahim AL-Naemi, MSc, Khadija Amin, MBBS, Yahya Ali Athlawy, Bsc, Akram Awad, BSc, and Zhonghua Sun, PHD

Abstract: Introduction: Basidiobolomycosis is an unusual fungal disease that rarely involves the visceral organs such as gastrointestinal tract. Gastrointestinal basidiobolomycosis (GIB) has been increasingly reported in the literature, and it is an emerging disease from arid regions worldwide, in particular, the south-western Saudi Arabia. We report a case of GIB in a 36-year-old Saudi Arabian male patient showing resistance to itraconazole and best treated with voriconazole. Computed tomography showed diffusely thickened small bowel with edematous change.

Conclusions: As GIB presents diagnostic challenges due to lack of specific features, this case emphasizes the importance of considering GIB in the differential diagnosis in patients presenting with fever, abdominal pain with fast-growing abdominal mass.

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Abbreviations: CT = computed tomography, ESR = erythrocyte sedimentation rate, GIB = Gastrointestinal basidiobolomycosis.

INTRODUCTION

Basidiobolomycosis is a rare fungal infection caused by *Basidiobolus ranarum*, a member of the subphylum Entomopthoromycotina, previously included in the Zygomycete class.[1] Visceral involvement by basidiobolomycosis is rare with the first case of gastrointestinal basidiobolomycosis (GIB) reported in 1964.[2] Only 73 cases of GIB have been reported in the medical literature so far.[3,4] The most common presenting symptom is abdominal pain, followed by fever, weight loss, or abdominal mass.[4]

Diagnosis of GIB is clinically difficult due to the fact that GIB occurs in immunocompetent patients and so it may not be clinically suspected.[4] Furthermore, its clinical presentation is nonspecific, with no identifiable risk factors. All age groups are susceptible to be infected, although the mean age was reported 37 years, according to a recent review.[4] Itraconazole, an antifungal agent is the most common drug used to treat GIB. We present a case of GIB showing resistance to itraconazole but susceptible to voriconazole, another effective antifungal agent but less commonly used in clinical practice. We emphasize the importance of considering GIB in differential diagnosis when dealing with patients presenting with fever and abdominal pain.

CASE REPORT

Written informed consent was obtained from the patient for the publication of this case and accompanying images.

A 36-year-old male patient living in the Gazan region of Saudi Arabia was admitted in the surgical unit of Khamis Mushayet hospital 1 year ago due to suspected appendicitis. During the surgery, a cecal mass was detected and right hemicolectomy with ileostomy was done. Histopathology revealed gastrointestinal basidiobolomycosis caused by *B. ranarum.* Antifungal drug itraconazole was given twice a day to the patient following the operation, but the patient's condition did not improve. It was unclear whether the infection caused by *B. ranarum* was resistant to itraconazole or the patient compliance was poor. The patient was on itraconazole for 6 months while he was still symptomatic. The decision of using the same medicine (itraconazole) was determined through consultation with the hospital's senior pharmacist and medical consultant with prolonged treatment with itraconazole considered the best option.

He was readmitted for chronic fever and abdominal pain for 6 months. We suspected intestinal obstruction. There was no history of vomiting, melena, or rectal bleeding. Apart from a temperature of 38.4°C and ileostomy in situ, there was leukocytosis (16000 mm^3), eosinophilia count (18%), and raised erythrocyte sedimentation rate (90 mm/L) (ESR). Other laboratory analyses such as urea nitrogen, serum creatinine, and liver function test were within normal range. Noncontrast and contrast-enhanced abdominal computed tomography (CT) scans showed generalized markedly thickened wall of the small bowel with edematous changes (Figure 1).

Voriconazole was planned for the patient in a dose of 200 mg (intravenously). The rationale for choosing voriconazole is that it is proved to be a very effective antifungal drug in the treatment of rare fungal infections or those infections showing resistance to other antifungal agents.[5,6] The treatment with voriconazole was continued for 2 weeks and the general condition of the patient improved. The fever subsided on the 5th day of commencement of treatment. Leukocyte (13300 mm^3) and eosinophil counts (9%) dropped after 1 week of initiation of voriconazole. The patient was discharged at the end of 2 weeks and orally taken voriconazole 200 mg twice a day was prescribed for 6 months. The patient was advised to attend the surgical outpatient clinic on a fortnightly basis to monitor the

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From the Department of General and Laparoscopic Surgery, Sabya General Hospital Jazan, 3261300, Kingdom of Saudi Arabia (AQA-N, LAK, IA-N, SA, YA-A, AA); and Department of Medical Radiation Sciences, Curtin University, Perth, 6845, Australia (ZS).
Correspondences: Awaji Qasim Al-naemi, Department of General and Laparoscopic Surgery, Sabya General Hospital Jazan, 3261300, Kingdom of Saudi Arabia (e-mail: awaji10@hotmail.com).
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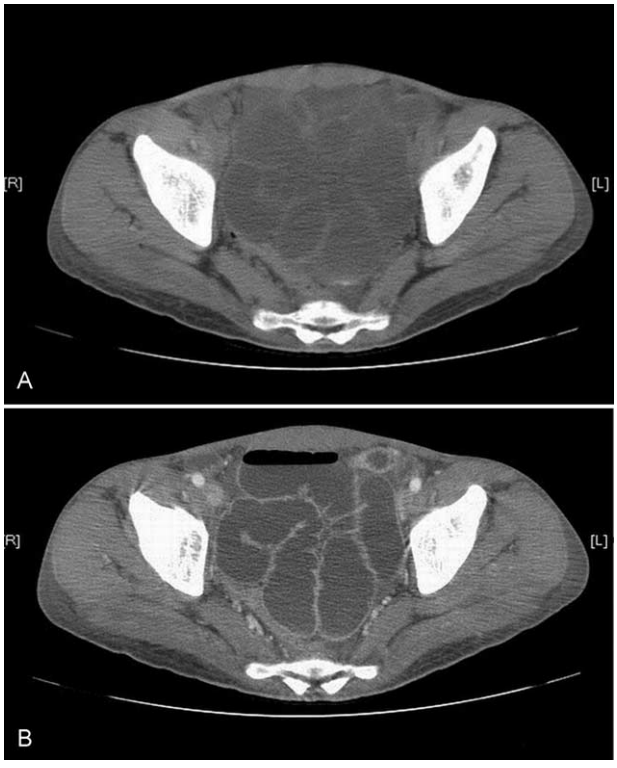

FIGURE 1. A: Non-contrast CT shows diffuse swelling of the small bowel. B: Contrast-enhanced CT reveals the wall thickening in the small bowel due to inflammatory changes. CT = computed tomography.

patient for potential complications such as blindness and cardiac ischemia associated with voriconazole. So far the patient was reviewed twice in the outpatient department with subsidence of the clinical symptoms and no complications.

DISCUSSION

Gastrointestinal basidiobolomycosis is an unusual fungal infection of the gastrointestinal tract caused by *B. ranarum*. It is rarely reported in the medical literature although sporadic cases have been reported worldwide, predominantly in the United States and Saudi Arabia, with some additional cases observed in Brazil and Iran.[4,7–10] Recently, this disease has become increasingly recognized with 73 cases reported in the literature.[3,4] Diagnosis of GIB is difficult as its occurrence in immunocompetent host is a cofounder. In addition, the nonspecific clinical presentations, such as fever, abdominal pain, or abdominal mass, could mimic infectious gastrointestinal tract disease or tumors.[11–14]

Flick et al reviewed the CT images of patients diagnosed with GIB and reported that colorectal mass is the most common finding, followed by hepatic mass or small bowel mass.[3] Bowel wall thickening was noticed in 25% of the reported cases, whereas bowel perforation or abscess was less commonly seen. The abdominal mass detected by CT in GIB patients is usually mistaken for neoplasm or inflammatory bowel disease. Similarly in our patient the initial diagnosis was appendicitis and the cecal mass was noticed only during the surgery. As suggested by Flick et al, GIB should be considered in patients presenting with a fast-growing abdominal mass. Another useful clue is the failure of nonantifungal medical treatment in such patients.

Surgical resection of the infected tissue coupled with prolonged treatment with itraconazole appears to be the best available clinical option. As patients treated with itraconazole showed complete resolution of the infection in most of the reported cases, Khan et al had questioned the use of surgical intervention.[15] Al Saleem et al avoided surgical intervention in the management of their case with patient showing excellent response to orally administered voriconazole.[14] They suggested that the option of choosing surgery is determined by the nature of the disease and its location, extension, and the patient's condition. It is well known that itraconazole is the most frequently used antifungal agent (73%), followed by amphotericin (22%), ketoconazole (8%), and voriconazole (5%).[4,7] Of these antifungal agents, voriconazole is a valuable and generally well-tolerated drug for the treatment of rare fungal infections and infections resistant to other antifungal agents.[5] Findings in this case report highlight the use of voriconazole as an alternative option in patients with no effective outcomes after being treated with other antifungal drugs.

In conclusion, we present a case of gastrointestinal basidiobolomycosis with abdominal pain and mass as the initial presenting symptoms. Although diagnosis is confirmed by histopathology after surgical removal of the mass in the gastrointestinal tract, the antifungal treatment using itraconazole is not effective. Use of voriconazole in this case shows significant improvement of the clinical symptoms. Findings of this case report could assist clinicians to better manage patients with gastrointestinal basidiobolomycosis with the aim of avoiding any complications associated with this rare disease.

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