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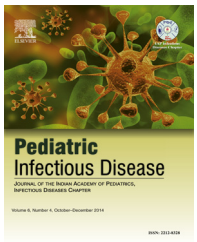

Case Report

# Pediatric gastrointestinal basidiobolomycosis

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ABSTRACT

Basidiobolomycosis is a rare fungal disease caused by *Basidiobolus ranarum*, which is an environmental saprophyte. It is a chronic inflammatory disease that is generally restricted to the subcutaneous tissue and rarely involves the gastrointestinal tract. With the intent to spread awareness of this potentially life threatening and rare infection, we report a 4-year-old boy presenting with abdominal pain and fever with eventual diagnosis of gastrointestinal basidiobolomycosis. We discuss the nonspecific and confusing symptoms of this rare infection and available treatment options in detail.



## 1. Introduction

Basidiobolomycosis ranarum is a rare known fungal organism, a member of the subphylum of Entomophthoramycotina.[1] Basidiobolus is an endemic infection in tropical and subtropical regions of the world, especially Africa, Latin America, and Asia.[2] The first human case of infection caused by *Basidiobolus ranarum* was one of subcutaneous mycosis, reported in 1956 in Indonesia, and other cases subsequently occurred in India, Africa, and South America.[3] Although the incidence of this infection is very low, recent studies have shown an emerging number of cases of visceral involvement due to *B. ranarum*.[4] In this article, we report a four-year-old boy with the eventual diagnosis of gastrointestinal basidiobolomycosis (GIB) from a southern province of Iran.

## 2. Case presentation

A 4-year-old boy from a southern province in Iran was referred to Bahrami Children hospital, a tertiary children's hospital, Tehran, Iran on December 2013 with chief complaint of abdominal pain and intermittent fever that first began approximately 45 days prior to his admission.

The very first symptoms of the patient began on September 2013, when he was admitted to a regional hospital because of abdominal pain. According to the documents of the mentioned hospital, in his physical examination, he was mildly dehydrated and ill. In his first abdominal examination, a generalized tenderness was detected. His vital signs were stable except for a temperature of 38 °C axillary. His past medical history was significant for a history of a unilateral undescend-

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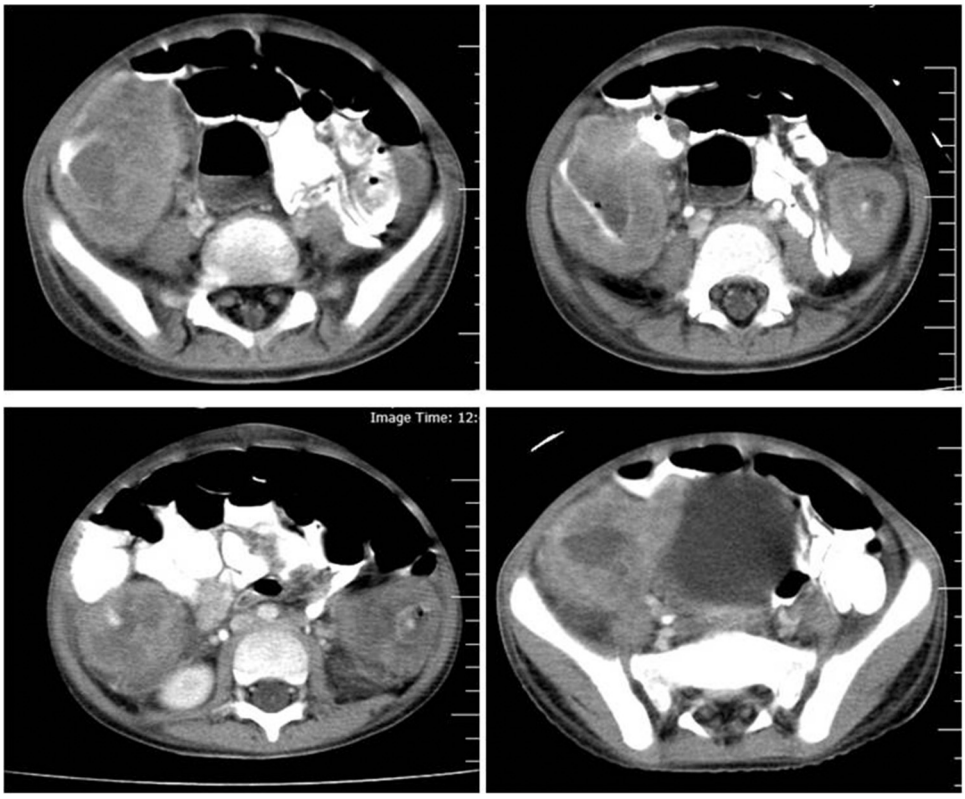


**Fig. 1 – Abdominopelvic contrast-enhanced CT scan of our patient. Two extensive heterogeneous masses in the terminal ileum, which were extended to the cecum.**

ed testis and cleft palate that was repaired surgically. He was otherwise a healthy boy.

He was diagnosed to suffer from cholelithiasis by ultrasonography. Unfortunately, he went through a laparoscopic surgery for cholecystectomy.

After the surgery, his abdominal pain and fever did not resolve. Intermittent bilious vomit was also added to his symptoms. The patient was referred to another hospital for further evaluation and management.

In the second hospital, during the physical exam, the patient appeared to be icteric and febrile. In his abdominal examination, a tender palpable mass in the right lower quadrant was discovered. Rectal examination was unremarkable. A repeated abdominal ultrasonography and abdominal computerized tomography (CT) scan were performed. Two extensive masses in the terminal ileum, which were extended to the cecum, were reported in the imaging process (Fig. 1).

The patient underwent a right hemicolectomy. During the surgery, three more masses were detected in the peritoneum. Histopathologic findings from the resected intestinal masses revealed extensive inflammation, granulomas with eosinophils and numerous fungal hyphae, and zygospores surrounded by eosinophilic material called the Splendore-Hoeppli phenomenon suggestive of basidioblomycosis infection. We unfortunately could not isolate the fungus from the intestinal specimen. Therefore, he was first treated with IV amphotericin B with 0.5 mg/kg stat and then 1 mg/kg/day, and when there was no significant response to treatment after about a week, it was changed to itraconazole capsules 10 mg/kg/day. After about ten days of unsuccessful treatment, the patient was referred to our hospital for further management.

On his admission in our hospital, the patient appeared ill and emaciated. In his first physical exam, his pulse rate, blood pressure, and temperature were normal. His abdominal examination revealed marked distention and a tender palpable mass in the left lower quadrant. An ileostomy, colostomy, and a bile drainage tube were noted in abdominal examination.

Results of laboratory investigations showed white cell count: 13,400/$mm^3$ (neutrophils 77% and lymphocyte 21%), hemoglobin: 11.5 mg/dl, platelet: 188,000/$mm^3$, ESR: 7, CRP: 57, AST: 67, ALT: 21, total bilirubin: 1.8 mg/dl, and direct bilirubin: 1.1 mg/dl. Electrolyte levels, blood urea nitrogen values, creatinine levels, and urinary analysis were normal.

Since his clinical symptoms persist and hematemesis and hematochezia were added to his symptoms, the antifungal treatment was changed to intravenous voriconazole 6 mg/kg/day BID for the first 24 h and then continued with 4 mg/kg BID. Because of his poor general condition, he was inoperable and no further surgical procedure could be performed. Unfortunately, though aggressive antifungal therapy was continued for the patient, his general condition deteriorated and he experienced multiorgan failure and expired on the 10th day of his admission in our hospital.