Gastrointestinal Basidiobolomycosis in Children

Khalid Shreef*

Department of Pediatric Surgery, Armed Forces Hospitals, Khamis Mushyet, Saudi Arabia

*Corresponding Author: Khalid Shreef, Department of Pediatric Surgery, Armed Forces Hospitals, Khamis Mushyet, Saudi Arabia.

Received: March 20, 2020; Published: April 17, 2020

Gastrointestinal Basidiobolomycosis (GIB) is an emerging fungal infection that manifests in the skin and rarely involves other systems. Visceral involvement by basidiobolomycosis is rare with only few cases (73 cases) have been reported worldwide so far. Most of the cases of pediatric GIB were reported from the southern region of Saudi Arabia. It is caused by Basidiobolus ranarum, which does not usually invade blood vessels and rarely disseminates. All age groups are susceptible to infection. According to a recent reviews, the mean age was 37 years. Male to female ratio is 5:1.

Basidiobolus ranarum (B. ranarum) is a fungus of the order entomophthorales causing chronic subcutaneous zygomycosis of worldwide environmental distribution. Human disease is concentrated in tropical and subtropical regions. It is endemic in Uganda and certain areas of Africa, India and other parts of Asia, but it is found worldwide, even in areas where the disease has not been known. B. Ranarum has been isolated from leaves and decaying plants in southern and northeastern states of the USA and the gastrointestinal tract of reptiles, fish, amphibians, horses, dogs and insectivorous bats. Occasionally the fungus has been isolated from insects. Thus, stagnant water in ponds inhabited by fish or amphibians could be an important source of this fungus. Use of ponds as a water source for bathing or toilet purposes or ingestion of food contaminated with soil or animal feces are the most likely routes of gastro-intestinal basidiobolomycosis (GIB) infection. No specific risk factors for GIB have been identified; however, prior ranitidine use and longer residence in endemic areas may contribute to the risk.

Although, anaemia, elevated inflammatory markers and absolute eosinophilia appears to be prominent laboratory features in GIB, still the diagnosis of GIB is often elusive due to many reasons. Firstly, the non-specific clinical presentation in immunocompetent patients. Abdominal pain is the most common presenting symptom (94.4%) followed by constipation and abdominal mass (83.3%, 77.8% respectively); fever is only present in 22.2% of cases. In rare cases the presenting symptoms may be bleeding per rectum due to involvement the rectal wall. Secondly, based on the radiographic appearance of an abdominal (usually colonic) mass with spread to surrounding organs, especially when associated with alteration in bowel habits, the disease often mimics malignancy or Crohn’s disease which may delay diagnosis. Thirdly, the infection usually involves the non-mucosal layers of the gastrointestinal tract so that endoscopic imaging and biopsy specimens may show nonspecific inflammation and neutrophilic infiltrate is rare. Therefore, GIB is an infection that leads to diagnostic confusion. From my point of view, I do emphasize that diagnosis of GIB requires high index of suspicion, awareness and consideration. The diagnosis might be suspected in the previously healthy children, especially those living in, or near, tropical areas and developed symptoms that may suggest the diagnosis and associated with abdominal masses and eosinophilia.

Because the diagnosis is usually equivocal, tissue biopsy is mandatory. Although culture or serology tests are required for definitive diagnosis, histologic analysis can provide a probable diagnosis of GIB. Surgical specimens should be inoculated soon after resection because it does not survive at 4°C. Sabouraud agar is an adequate medium, and visible growth is usually present 2 to 3 days after incubation at 25°C to 30°C. Diagnosis is based on histopathology. The mold’s structural elements include both hyphae and zygospores. Typical mor-
Gastrointestinal Basidiobolomycosis in Children

Phologic features include hyphae that are irregularly branched, thin walled, occasionally septated and surrounded by a thick eosinophilic cuff (Splendore-Hoeppli phenomenon).

The current experience of treating patients with GIB is limited. Surgical resection of the infected tissue followed by prolonged antifungal therapy (more than 3 months) were the most commonly used options. However, fewer literatures had questioned the role of surgery in management of GIB. They successfully used antifungal therapy alone in the management of their case. Nevertheless, the long term prognosis in such cases remains unclear, and the obtaining tissue biopsy by interventional radiology, endoscopy or laparoscopy is usually associated with high risk of GIT perforation. From my experience, I do suggest that if the obtaining of tissue biopsy is hazardous or impossible then surgery is mandatory in such cases. If a patient is fit for surgery and the surgical resection is safe, then the aim is only resection of the affected bowel segments without mutilation. Surgery would provide a good specimen for histopathology and remove the nidus of infection thus reducing the duration of antifungal therapy with less drug side effects.

The best choice of antifungal agent is not clear, but many literatures. From my experience I suggested that itraconazole seems to be reasonable; it showed complete resolution of the infection in most of the reported cases. Voriconazole could be an alternative antifungal therapy for cases that have resistance to itraconazole. The efficacy of amphotericin B in visceral infections has been unsatisfactory, with resistance observed in greater than 50% of cases. Potassium iodide has been used successfully for treatment of subcutaneous basidiobolomycosis but not GIB.

To recap, GIB is an emerging infection in children that leads to diagnostic confusion. Diagnosis of this disease requires high index of suspicion, awareness and consideration of its possibility in the differential diagnosis in patients with abdominal masses and eosinophilia, in particular in endemic areas. Increased awareness of this clinical entity, early surgical resection of the infected tissue and prolonged treatment with Itraconazole offer the best chance for curing the disease.

Volume 7 Issue 5 May 2020
© All rights reserved by Khalid Shreef.