Gastrointestinal Basidiomycosis: An Unusual Fungal Disease?

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Abstract

Background: Basidiobolus ranarum is a fungus that usually causes subcutaneous infection. Medical literature rarely report gastrointestinal involvement, especially in tropical climate areas.

Case Report: Here we report a case of gastrointestinal basidiomycosis in an immunocompetent 43-year-old man from south of Iran who presented with abdominal pain, loss of appetite, and nausea. He had history of previous laparotomy 10 years ago, because of perforated peptic ulcer. He underwent an exploratory laparotomy. We found a mass in ascending colon; so, right hemicolectomy were done. Histologic founding were amazing. Eosinophilic sheath surrounding hyphae-like structures was seen. The diagnosis of basidiomycosis was established, so we prescribed antifungal agents for the patient. He expired 6 months later according to disseminated disease.

Conclusions: Gastrointestinal basidiomycosis is a rare and invasive fungal infection that imitates malignant tumors, inflammatory bowel disease, or even phlegmon of appendicitis. Diagnosis of gastrointestinal basidiomycosis needs a high index of suspicion. The physicians should be aware of this disease as differential diagnosis in tropical areas. Surgical resection and prolonged antifungal therapy is recommended, but in some cases, the disease may spread and cause death.

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Introduction

Basidiobolus ranarum is a fungus that can be found in soil, decaying vegetable, gastrointestinal track of amphibian, reptiles, and fish, and is endemic in tropical and subtropical regions. Consumption of contaminated fruit vegetables, unintentional ingestion of contaminated soil, gardening, and farming are maybe risk factors. Host factors such as gastric acid suppression (like our case), gastrectomy, and diabetes mellitus may also contribute to risk (1). It usually causes slowly progressive subcutaneous infection in trunk and limbs, not only in immunodeficient patients but also in immunocompetent patients. It rarely affects gastrointestinal track. Gastrointestinal basidiomycosis usually imitates inflammatory bowel disease, malignancies, appendicitis, or diverticulitis (2). Some cases are reported from United States and Middle East countries like Iran, Saudi Arabia, Kuwait, and Oman (3-13). In most cases the diagnosis were revealed after surgical resection.

Case Report

A 41-year-old man who lived in a small village in south of Iran with tropical climate, was admitted to emergency ward with periumbilical pain, nausea, loss of appetite, and malaise from two weeks ago. The pain was constant. He did not have change of bowel habit. He had history of upper midline laparotomy 10 years ago because of perforated peptic ulcer. His physical examination showed stable hemodynamic, and periumbilical and right lower quadrant tenderness with mild rebound tenderness, but no signs of peritonitis. Laboratory findings showed leukocytosis with eosinophilia, white blood cell (WBC): 21000/mm³, neutrophil: 69.2%, and eosinophil: 20.6%. Abdominopelvic sonography reported a heterogeneous large mass (123 × 73mm) at right lower quadrant that could be due to phlegmon. Computed tomography (CT) scan showed mucosal thickening in large bowel (Figure 1).

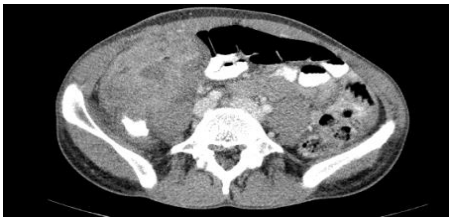

Figure 1. Computed tomography (CT) scan of abdomen, a large heterogeneous mass in right iliac fossa with an irregular thickened bowel wall

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Differential Diagnosis at this point included complicated (phlegmon of) appendicitis and colon cancer. We started broad spectrum antibiotics to manage the phlegmon of appendicitis, and we planned to do delayed appendectomy 6-8 weeks later. After a week, the sign and symptom of patient were not disappeared, and his abdominal pain was progressed. So, we planned laparotomy for him. The tumor was located in cecum, and regional lymph nodes were enlarged (Figure 2).

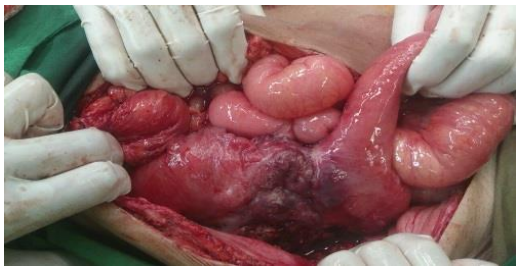

Figure 2. After midline laparotomy, we found a big cecal mass with enlarged lymph nodes.

The patient underwent right hemicolectomy (Figure 3).

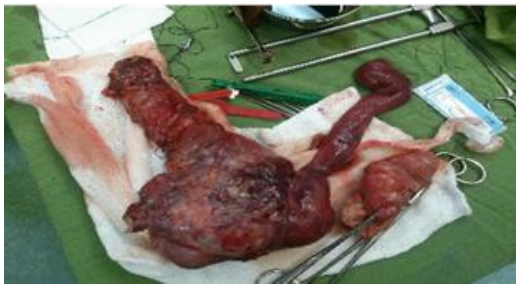

Figure 3. Right hemicolectomy specimen

Histopathologic study showed mucosa was edematous, and a 5 × 4 × 3 cm soft constant and creamy color mass was protruded in lumen and extended to the serosal layer (Figure 4).

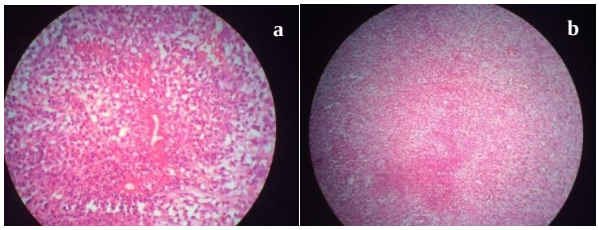

Figure 4. Granulomatous inflammation of colonic wall and periodic acid-schiff stain show fungal hyphae, hematoxylin and eosin 400x (a) and 100x (b).

Then, we prescribed amphotricin B and itraconazole for him. After 2 weeks from his laparotomy, he was discharged from hospital. Six months after, he was admitted in another hospital with an acute severe septic shock, and the patient expired.

Discussion

Basidiomycosis is a fungal infection that rarely affects gastrointestinal track, not only in immunodeficient patients but also in immunocompetent patients. Most of the cases are reported from tropical climate areas such as Arizona State in United States, India, Iran, and other countries around Persian Gulf like Oman and Saudi Arabia (3-13). There are no specific risk factors, and all age groups are at risk (2). It seems that basidiomycosis affected males more than females (3-5).

In a study in Iran, 14 cases of gastrointestinal basidiomycosis were reported. Patients had abdominal pain, fever, gastrointestinal mass, eosinophilia, and high erythrocyte sedimentation rate (ESR). All of their diagnosis were after surgery by histopathological findings (6).

Another study of 44 patients (range 2-81 year) with gastrointestinal basidiomycosis, presented that most of the patients were from United States (43%) and Saudi Arabia (25%). 64% were previously healthy. In their histories, common chronic medical disease were diabetes mellitus and gastric disorders. Colon was involved in 82%, small intestine in 36%, and liver and gall bladder in 30% (1). In another study in Saudi Arabia, 9 pediatric patients were reported with gastrointestinal basidiomycosis during 10 years (5).

5 patients with basidiomycosis from Saudi Arabia were misdiagnosed as cancers like lymphoma and carcinoma, tuberculosis, or inflammatory bowel disease (7). Diagnosis of gastrointestinal basidiomycosis is difficult as it is a rare condition, and there are not definite risk factors and no specific sign and symptoms. Of course it needs a high index of suspicion. The physicians should be aware of this disease as differential diagnosis in tropical areas. A 12-year-old Iranian boy was reported with basidiomycosis in left colon that mimic amebiasis by bloody diarrhea and abdominal pain (8). 7 patients, including 4 patients from Saudi Arabia, 2 from Iran, and 1 from Mali were reported separately with basidiomycosis in ascending colon with differential diagnosis of appendicitis and colon cancer. All underwent right hemicolectomy and treatment were accompanied with antifungal agents. One of them died because of septic shock (9-13).

Review of literature demonstrates increasing gastrointestinal basidiomycosis cases in immunocompetent patients in last two decades, especially in United States and Middle East (3). Surgical resection and antifungal agents for a long time is recommended in most cases. Result of monotherapy with amphotericin B was undesirable, as 50% of cases were resistant to this antibiotic (14). Combination of amphotericin B and an azole agent like itraconazole, ketoconazole or voriconazole for at least one year had the best results (1,5,9-11,14,15).

Conclusion

Basidiomycosis is a fungal infection that rarely affects gastrointestinal track. Signs and symptoms imitate malignant tumors, inflammatory bowel disease, or even phlegmon of appendicitis. It should be considered as a differential diagnosis in tropical climate. Despite the surgical resection and antifungal therapy, in some cases the disease may spread and cause death. We do not know much about risk factors, early detection cues, and the best and definite treatment; so it requires more studies to answer the questions.

Conflict of Interests

Authors have no conflict of interests.

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