Isolated Hepatic Basidiobolomycosis in a 2-Year-Old Girl: The First Case Report

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1. Introduction

Zygomycosis includes 2 orders, one of which causes fungal infections in an immunocompromised host (Mucorales) and the other in an immunocompetent host (Entomophthorales) (1).

Basidiobolus ranarum belongs to the second group and is a saprophyte found mostly in soil and decaying vegetable material (2). It is a low virulent fungus, and the first human case of this fungal infection was reported in 1956 in subcutaneous tissue (3). Since then, many cases of subcutaneous basidiobolomycosis have been reported. However, the gastrointestinal (GI) involvement of this fungus is an emerging infection and has rarely been reported (4). Recently, a few cases of GI basidiobolomycosis, accompanied by liver involvement as part of a disseminated disease, have been reported (5).

To the best of our knowledge, no case has been reported in the English literature with an isolated liver mass caused by basidiobolomycosis without the involvement of any other organ. Accordingly, herein we report our experience with a 2-year-old girl, who presented with a liver mass subsequently identified as basidiobolomycosis.

2. Case Presentation

A 2-year-old well-nourished and well-developed girl from the Iranian city of Kangan (Bushehr province) presented with vague and generalized abdominal pain. She was the first child of the family, born via normal vaginal delivery without any specific disorder. She had had a normal infancy until 2 months prior to her referral, when she developed abdominal pain with no response to routine treatment.

Physical examination was normal, except for mild hepatomegaly. Laboratory tests showed microcytic hypochromic anemia (hemoglobin level = 7.8 gr/dL). White blood cell count was high (about 11–12000/cc) with significant eosinophilia (25% - 35%). Immunoserology tests revealed high C-reactive protein (CRP = 13 mg/L, normal < 6) and erythrocyte sedimentation rate (ESR = 83, normal for the patient's age < 10). Liver function tests showed high aspartate aminotransferase and alanine aminotransferase (57 and 45 IU/L respectively, normal < 31) and alkaline phosphatase (4030 IU/L, normal < 300). Stool occult blood was performed 3 times, and the results were all negative.

Abdominal ultrasonography demonstrated a prominent liver with a well-defined mass lesion measuring 40 × 35 cm. Another mass was detected in the hilar area. The other parts of the GI tract, including the stomach and intestine walls, were normal. Upper abdominal magnetic resonance imaging (MRI) showed normal thickness of the GI tract with no mass, but there were multiple masses in the liver. The first impression of both sonography and computed tomography scan was liver or biliary abscesses (Figure 1). Tru-Cut needle biopsy displayed a mainly normal liver with foci of eosinophil infiltration, which was nondiagnostic.
Therefore, the patient underwent surgery, which showed multiple nonencapsulated liver masses with ill-defined borders, the main one in the parenchyma and the other in the hilar area. During surgery, precise search was made to find any accompanied GI mass, but no mass was identified. Also, the omentum was completely free of any tumor or mass lesion. The masses were resected and sent for culture and pathologic studies. The pathology sections showed Splendore-Hoeppli bodies and many eosinophils as well as radiating eosinophilic granular material surrounding the fungal elements within the liver parenchyma and in the hilar mass within the lymph node tissue (Figures 2A and 2B). The fungal elements exhibited broad hyphae with thin walls with no septae or sparse septation.

According to the characteristic pathologic features, the diagnosis of hepatic basidiobolomycosis was made. However, all the cultures including fungal and bacterial were negative. The immune system, cellular and humoral, of the patient was thoroughly investigated, even for the possibility of chronic granulomatous disease. All of the studies regarding the immune system were normal.

Figure 1. A, B: Magnetic Resonance Imaging of the Abdomen Shows Multiple Low-Signal Masses in the Liver Associated With Biliary Dilatation

Figure 2. A, B: Degenerated Fungal Hyphae Surrounded by Granulomatous Reaction and Many Eosinophils in the Liver (2a: Low Power, 2b: High Power)
the possibility of a hepatic abscess was also considered. In most of the previous cases of GI basidiobolomycosis, the final diagnosis was made after surgery and resection of the liver mass (9). Nonetheless, the gold standard for the diagnosis of every fungal infection is culture. In the majority of the previously reported cases of GI basidiobolomycosis, culture was either negative or was not performed because of the unavailability of the proper tissue (5). In our case, cultures turned out to be negative. The pathologic characteristics of this fungal infection are the presence of Splendore-Hoepli bodies with many eosinophils and degenerated fungal hyphae (13). It can cause liver granuloma with heavy infiltration of eosinophilic liver granuloma and should be considered in the differential diagnosis of hepatic granulomas (15).

The best treatment in this pathogen is the resection of the mass, accompanied by antifungal therapy including itraconazole or amphotericin B. Our patient showed a dramatic response to amphotericin B. In less than a week, eosinophilia disappeared and ESR returned to normal and within 2 weeks, she resumed weight gain and her abdominal pain subsided. She is still under treatment, and the plan is to continue the antifungal therapy for at least 6 months, because our previous experience showed the high possibility of recurrence after an early discontinuation of the treatment.

In conclusion, basidiobolomycosis should be considered as a differential diagnosis of hepatic abscess with or without GI involvement to prevent delayed treatment.

### Authors’ Contributions

Bita Geramizadeh diagnosed the case and wrote and prepared the paper. Anahita Sanai Dashti handle the patient. Mohammad Rahim Kadivar treated the patient. Shirin Kord assisted in data collection.

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