Gastrointestinal Basidiobolomycosis
First case report from Oman and literature review

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**Abstract:** Gastrointestinal basidiobolomycosis (GIB) is a rare fungal infection with few reported cases worldwide. We report here the first case diagnosed in Oman in a previously healthy 5-year-old Omani female child who had been thought initially to have an abdominal malignancy. The case was referred to the Royal Hospital, Muscat, Oman, in July 2012. She was treated successfully with surgical resection and prolonged antifungal therapy (voriconazole). Physicians, including clinicians, radiologists and pathologists, should have a high index of suspicion for GIB when a patient presents with an abdominal mass and fever.

**Keywords:** Mycoses; Zygomycosis; Entomophthorales; Gastrointestinal Diseases; Child; Case Report; Oman.

Basidiobolomycosis is a known fungal infection of the skin and soft tissue in otherwise healthy individuals. Gastrointestinal basidiobolomycosis (GIB) is an uncommon infection in childhood. Few cases have been reported in the literature.†‡ The disease commonly manifests with fever, nausea, vomiting, abdominal pain, diarrhoea and/or an abdominal mass. Its non-specific presentation and unknown risk factors make it difficult to diagnose. In most reported cases the initial diagnosis is malignant neoplasm, tuberculosis or inflammatory bowel disease.†‡§

A definite diagnosis of basidiobolomycosis requires a microbial culture of *Basidiobolus ranarum* from fresh aspiration or surgical specimens. Histopathological examinations reveal a characteristic appearance of the culture.§ A favourable outcome depends on early diagnosis, the institution of appropriate antifungal therapy and surgical debulking.¹

In the Middle East, the largest series of paediatric GIB cases was seen in Saudi Arabia, with 11 reported in 2012.¹² This paper reports the first case of laboratory-confirmed abdominal basidiobolomycosis in a child from Oman.

**Case Report**

A 5-year-old Omani female was referred to the Royal Hospital, Muscat, Oman, in July 2012 with a two-week history of nausea, vomiting, abdominal pain and low-grade fever. The child had been diagnosed with acute appendicitis in a regional hospital and had had an appendectomy. The intraoperative findings suggested a normal appendix with a paracaelic mass. The subsequent histopathological examination also showed that the appendix and regional lymph nodes had no significant morphological abnormalities. The postoperative course was eventful with high fever and the slow return of bowel functions. An ultrasound of the abdomen revealed a hypoechoic circumferential mural thickening involving the caecum and ascending colon with luminal effacement suggestive of colitis or typhilitis. A computed tomography (CT) scan of the abdomen showed a large mass of heterogeneous

**References:**


density in the right iliac fossa inseparable from the bowel loops, with a markedly thickened irregular bowel wall involving the caecum and ascending colon and, to a lesser extent, the terminal ileum and, to a lesser extent, the terminal ileum, probably suggestive of non-Hodgkins lymphoma, typhilitis or adenocarcinoma [Figure 1].

The parents noticed that the child had lost a significant amount of weight (2.5 Kg) in a month even though she had previously been healthy and had had no blood-stained stools, constipation or pica. She had no history of recent travel abroad or contact with sick people. The child attended school regularly and had no history of previous hospital admissions. The family, who lived in the Al-Dakhiliyah region of Oman, an area with many arable farms surrounded by large areas of desert, used the government water supply for drinking. Her past medical and family histories were insignificant and she was fully immunised for her age.

The physical examination on admission revealed a sick, febrile child with a few palpable discrete and non-tender lymph nodes in the neck. No skin rashes or oral thrush was noted. The abdomen was soft with a tender large mass in the right iliac fossa. The liver and spleen were not palpable.

At this stage, the differential diagnoses were abdominal malignancy, lymphoma, inflammatory bowel disease, tuberculosis or infected fluid collection. Hence, an urgent ultrasound-guided aspiration and biopsy of the abdominal mass were done. The aspiration showed purulent fluid which was sent for both culture and cytological examinations. In addition, a true-cut biopsy specimen was sent for histological examination. The child was then started on piperacillin-tazobactam, gentamicin and metronidazole.

The complete blood count (CBC) highlighted a remarkable increase in eosinophils (6.2 x 10^9/L) with a total white blood count (WBC) of 14.2 x 10^9/L.

The peripheral blood film showed no significant morphological abnormalities other than eosinophilia. Her erythrocyte sedimentation rate, lactate dehydrogenase amounts, uric acid levels and chest X-ray were all normal.

On the third day of admission, the cytology report of the mass aspirate showed fungal filaments resembling mucormycosis. Liposomal amphotericin B was added to the existing course of antibiotics. She continued with intermittent high-grade fevers up to 39°C. She also had nausea and colicky abdominal pain which were suggestive of a partial obstruction.

The patient underwent an exploratory laparotomy which revealed a large, inflamed, necrotic mass arising from the caecum and ascending colon. The mesentery adjacent to the mass was found to be thickened. A small ulceration was seen in the colonic mucosa close to the resection margin. A few slightly enlarged mesenteric lymph nodes were present. The liver and spleen appeared normal. A right hemicolectomy was performed and the bowel continuity was restored by an ileocolic anastomosis.

Posaconazole was added to the antifungal regimen and after two days the patient’s temperature started to lower and her abdominal pain improved significantly. The surgical wound still showed redness and induration despite the child being on liposomal amphotericin B and posaconazole. Therefore, she underwent extensive debridement of the abdominal wall and swabs were sent for bacterial and fungal cultures.

The histopathological examination of the resected mass showed numerous scattered broad pauciseptate fungal hyphae embedded in eosinophilic material rich in eosinophils and some neutrophils representing the Splendore-Hoeppli phenomenon [Figure 2]. The special stains showed no acid fast bacilli and there was no evidence of malignancy.

A subculture on Sabouraud’s dextrose agar yielded Basidiobolus spp. On the third day of inoculation, the...
colonies were initially white and radially-folded with short aerial hyphae. With age, the colonies turned cream to yellow-brown in colour. The microscopic examination revealed pauciseptated hyphae measuring 5–20 μm wide. Some of the hyphae fragmented into short hyphal bodies. Zygospores with two beak-shaped remnants of the copulation tube established this organism as Basidiobolus spp. [Figures 3 A and B]. Another sample from the infected surgical wound also grew the Basidiobolus species.

The antifungal therapy was modified after isolating the Basidiobolus spp. Liposomal amphotericin B and posaconazole were discontinued and intravenous voriconazole was started at a dose of 8–15 mg/Kg/day, divided into two 12-hourly doses. The abdominal wound was closed when repeated cultures showed no fungal growth. On the 28th day, the patient was discharged with a three-month course of oral voriconazole (15 mg/Kg/day, divided into two 12-hourly doses).

When she visited the outpatient clinic a month later, the patient showed a dramatic improvement in her general condition. She had started gaining weight and the wound was healing well. Her CBC showed a normal eosinophil count and the abdominal ultrasound showed no masses or thickening of the bowel. It was decided to keep her on antifungal treatment for a year with follow-up visits at three-month intervals. It was also decided to perform abdominal CT/magnetic resonance imaging scans a week prior to the end of therapy, or sooner if she became symptomatic again.

**Discussion**

Zygomycosis is caused by the Mucorales and Entomophthorales orders of fungi. Entomophthoromycosis consists of both basidiobolomycosis and conidiobolomycosis. Basidiobolus ranarum, as a causative species of basidiobolomycosis, was first isolated in 1955 in the USA from decaying plants and was later found in soil and vegetation worldwide. It can be found in the faeces of amphibians, reptiles and insectivorous bats and is known to cause skin and soft tissue infections in otherwise healthy individuals. Recently, basidiobolomycosis has emerged primarily as a systemic infection of the alimentary tract; however, as it generally does not invade the blood vessels, it rarely disseminates.

Vikram et al. reported the worldwide occurrence of GIB between 1964 and 2010 as 44 cases. Of those, a total of 19 cases were reported from the USA, of which 17 were from Arizona alone. A probable environmental link to the desert climate was suggested. Saudi Arabia had the second highest prevalence with a total of 11 cases. The youngest patient was 2 years old and the oldest 81 years old. The majority of patients presented with abdominal pain, an abdominal mass, fever, weight loss, constipation and/or diarrhoea. Most of the patients had peripheral eosinophilia (76%) and the culture was positive in 71% of the cases. The colon and rectum were the most commonly involved organs, but it can also infect the small bowel, liver and stomach.

Predicting potential risk factors that predispose a person to GIB is limited in these different case series by the small number of patients, incomplete patient information and variations in the follow-up period. A six-patient case-control study from 2001 suggested that male gender, a history of diabetes mellitus, peptic ulcer disease and pica could be risk factors, but these were hard to confirm in a later larger cohort.

On histopathological examination, the fungal hyphae are irregularly branched, thin-walled and occasionally septate. In most reported cases, the fungal hyphae were surrounded by a thick eosinophilic cuff—the Splendore-Hoepli phenomenon. This

**Figure 3 A & B:** Microscopic morphology of Basidiobolus spp showing globose one-celled conidia that are forcibly discharged from a sporophore (arrow) at x 100 magnification with lactophenol blue stain (A). Microscopic morphology of Basidiobolus spp showing numerous round smooth thick-walled zygospores (arrow) at x 100 magnification (B).
phenomenon is not specific to GIB and can also be seen around other fungi, helminths or their ova, bacterial colonies or, rarely, around suture material in tissues.3

The treatment for GIB combines both surgical resection and appropriate antifungal therapy. The possibility of eradicating the infection using only antifungal therapy without surgical resection remains theoretical as there have not been many opportunities to follow this treatment method. The importance of early surgical intervention for paediatric GIB was emphasised in a retrospective review of nine paediatric cases managed at the King Faisal Specialist Hospital & Research Centre, Riyadh, Saudi Arabia.13

The azole antifungal group is considered the best for treating GIB, including ketoconazole,itraconazole and voriconazole. There is concern about the resistance of B. ranarum to amphotericin B products and some reported mortality in patients treated with them.10,14

Almost all previously reported cases were initially misdiagnosed as inflammatory bowel disease, malignancy, diverticulitis, appendicitis and gastrointestinal tuberculosis, among others.1,3,9,15 In the current case, the patient was initially thought to have appendicitis and then was referred to the Royal Hospital in order to rule out lymphoma. Limited awareness about the histopathological findings in such an infection led to a misleading report that suggested mucormycosis in the patient. This resulted in the delay in instituting the proper antifungal course of treatment earlier in the course of the illness.

The previously healthy child reported in this report presented with abdominal pain, abdominal mass, fever and weight loss. She had caecal and colonic involvement. Her CBC showed peripheral eosinophilia. The histopathology of the biopsy showed typical fungal hyphae surrounded by the Splendore-Hoeplli phenomenon, which together with the clinical input met the definition of probable GIB. The fungal culture later confirmed the diagnosis. This case highlights the crucial importance of performing appropriate cultures to confirm the diagnosis of GIB. She had a successful outcome with surgical resection and prolonged antifungal treatment. This is the first reported and diagnosed case of GIB in Oman; however, some cases may have been missed previously, especially those with atypical presentations and/or no request for microbial cultures.

Conclusion

GIB is an invasive fungal infection that should be among the differential diagnoses for children presenting with abdominal masses and fever. An awareness of this condition among paediatricians, paediatric surgeons and pathologists may lead to the discovery of more cases and help the treating physician to arrive at a diagnosis earlier and manage the case appropriately. A worldwide collection of case studies is needed to identify risk factors for GIB.

References