INTRODUCTION
Basidiobolomycosis is a disease caused by saprophytic fungus called Basidiobolus rararum (B. rarum). It is known to cause chronic dermatological manifestations but rarely produces systemic disease which sometimes becomes very serious and fatal, if timely management is not being taken. The habitat of basidiobolus rararum is soil, decaying organic matter and gastrointestinal tract of some animals. It mainly affects those living in the tropical regions with humid and hot environment. The clinical presentation of this indolent disease is similar to many common conditions involving gastrointestinal tract like infections, inflammatory bowel disease and infiltrative conditions.

We report here a case of Saudi boy who had a very stormy course of disease before diagnosis of basidiobolomycosis could be made, but treated successfully.

CASE REPORT
A 10-year Saudi boy was admitted with one-year history of recurrent abdominal pain associated with fever and weight loss. He had appendectomy for presumed acute appendicitis at the beginning of his disease at a local hospital. Since then he had recurrent course of abdominal pain and fever with multiple admissions in different hospitals, for which he received various antibiotics and antipyretics. Subsequently, he was diagnosed as Crohn's disease after upper gastro-intestinal endoscopy and colonoscopy. Despite being on appropriate treatment for CD including steroids, 5-Aminosalicylic acid and azathioprine with good compliance, his symptoms never resolved. The family denied any history of diarrhea, constipation or bleeding per rectum; however, he had significant weight loss (approximately 7-8 kg) during one-year period. Family history of inflammatory bowel disease (IBD), immunodeficiency or malignancy was negative. His growth percentiles were within normal limits (weight 34kg and height 137cm, both above 50th centile) with unremarkable systemic examination except tenderness in right iliac region. His digital rectal examination was also unremarkable.

His investigations showed persistently raised inflammatory markers with erythrocyte sedimentation rate (ESR) of 105mm/h and significant peripheral eosinophilia (range 15-44%) and radiological evidence of mucosal wall thickening at the ileocecal region on two abdominal computerized tomography (CT) scans as shown in the Figure 1. Repeat colonoscopy revealed marked inflammatory changes in the caecum and terminal ileum along with pseudopolyps formation (Figure 2) with normal rest of the colon. Histopathology report depicted mild chronic inflammation with normal crypts structure and suggestive of non-specific colitis. Moreover, the biopsy tissue revealed no positivity for tuberculosis and stains negative for fungus. Although, histopathology was not supportive of IBD, based on clinical history, raised inflammatory markers, endoscopic findings, he was initiated on the treatment of Crohn's disease and discharged home on tapering steroids, azathioprine and elemental diet (exclusive polymeric formula).

CASE REPORT
Ileocolonic Basidiobolomycosis in a Child: An Unusual Fungal Infection
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ABSTRACT
Systemic basidiobolomycosis is a rare fungal infection caused by Basidiobolus rararum (B. rarum). The clinical presentation is non-specific and is similar to many gastrointestinal conditions such as Crohn's disease (CD). The most consistent findings of basidiobolomycosis are recurrent abdominal pain, weight loss, fever and peripheral eosinophilia. Most of the patients are diagnosed on surgical resection of the involved region along with compatible histopathological findings like transmural inflammation, granulomas with eosinophilic infiltration (Splendore-Hoeppli phenomenon) and more specifically detection of fungal hyphae on fungal stains. Effective and curative treatment for systemic basidiobolomycosis is available, if diagnosed and managed properly in time. We report here a Saudi boy who had ileo-caecal basidiobolomycosis, but diagnosed after a prolonged course of illness.

Key Words: Basidiobolomycosis. Child. Ileocecal infection.

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Three weeks later, he was brought again to the emergency room with abdominal pain, fever and bilious vomiting, and no bowel activity for three days. He appeared to be sick, dehydrated and febrile. Now, there was a mass in the right iliac fossa and the abdomen was diffusely tender and tense with guarding. The child was taken to theater for emergency laparotomy for suspected intestinal obstruction. Intraoperatively, there was a firm abdominal mass arising from cecum and adhered to anterior and lateral abdominal wall with a retroperitoneal attachment and fibrosis. The mass was resected with right hemicolectomy and ileo-colonic anastomosis was made. The histopathological analyses of the mass showed extensive mural inflammation with necrotizing granulomas and positive for fungal organisms consistent with basidiobolomycosis as shown in Figure 3. Intravenous anti-fungal voriconazole 7mg/kg twice daily was commenced in addition to total parental nutrition (TPN) and treatment for CD discontinued. Unfortunately, he had complication on 7th postoperative day with the intestinal leak at the anastomotic site. He underwent for a second emergency laparotomy for correction of the leak and formation of ileostomy and mucous fistula. Afterwards, he had smooth course and was discharged on oral voriconazole after 6 weeks of intravenous infusion. At 3- and 6-month follow-up, he was doing well and regained weight by 4 kgs. He was continued with oral voriconazole by the infectious diseases department.

**DISCUSSION**

Systemic basidiobolomycosis is a rare fungal infection and very few case reports and series are published in the literature. The majority of patients with systemic disease were reported from Saudi Arabia, Iran, Brazil, Nigeria and United States.2-5 Residents of tropical and sub-tropical areas are at greater risk where this fungus finds favorable environment for growth.2 Males outnumber females and it is still undetermined why this gender predilection is present in this disease. It is reported in all the pediatric age group and youngest child was of 13 months from Iran (age range 13 months-16 years).6 Various modes of transmission are described including ingestion of soil, contaminated food with fungus or animal feces. The usual gastrointestinal presentation includes recurrent abdominal pain, fever and weight loss which are similar to our case. Bloody diarrhea and mass in the right iliac fossa were also reported in the literature. Other systemic manifestations depend on the site of involvement and include vomiting, bloody diarrhea, jaundice or urinary symptoms and fistulous tracts. Abdominal pain, weight loss, fever and peripheral eosinophilia were the consistent gastrointestinal findings in most of the patients reported in different studies, similar to this case.6,7

The major differentials of basidiobolomycosis in children are Crohn's disease, infections like amoebiasis, intestinal tuberculosis, appendicitis or appendicular mass, sarcoidosis and malignancies. Crohn's disease is a big mimicker of basidiobolomycosis because of similar presentation and majority of previously reported patients were managed as IBD or appendicitis similar to this case.3,4,7
There are neither specific clinical symptoms nor investigational criteria set for the diagnosis of these children because of the rarity and non-specific symptoms. Only one patient was diagnosed, based on endoscopic biopsies and rest had confirmation only when the mass was resected and examined microscopically. It might be the reason that B. rararum affects the submucosal deeper tissues more than the mucosa; and endoscopic mucosal biopsies do not provide sufficient material for identification, as happened in this case. The surgical specimens have good yield for identification of B. rararum with evidence of granulomas and inflammation and also facilitates culture positivity. There are no consensus guidelines for optimal management strategies. In addition to surgical intervention that is required for most of the patients, long term anti-fungal therapy is indicated to fully eradicate the disease. Pharmacological options include amphotericin B, itraconazole, voriconazole and posaconazole either in combination or monotherapy. Most studies showed good response to amphotericin B with itraconazole given for 6 months to 1 year, but recent evidence of monotherapy with voriconazole is also remarkable. Duration of therapy is not known and it is also yet undetermined if surgical resection is being done, for how long the therapy should continue. The long-term outcomes for these children are excellent, if picked and managed early. Previous studies showed high mortality in the younger age group and particularly if diagnosis is delayed. Invasive basidiobolomycosis carries a high morbidity and mortality, if not diagnosed and managed early. Unusual course of the disease and inappropriate response to common gastrointestinal mimickers shall lead to think about basidiobolomycosis.

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