CASE REPORT/CAS CLINIQUE

Chronic rhinofacial basidiobolomycosis caused by Basidiobolus ranarum: Report of a case from Aseer Region, Kingdom of Saudi Arabia

Rhinofacial basidiobolomycosis chronique causé par Basidiobolus ranarum : rapport d’un cas de la région Aseer, Royaume de l’Arabie Saoudite


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Summary We present a case of fungal sinusitis caused by Basidiobolus ranarum in a 22-year-old male patient with chronic rhinosinusitis in Aseer region, Kingdom of Saudi Arabia. The patient was admitted with nasal obstruction accompanied by itching, sneezing, rhinorrhea, epistaxis and recurrent headache. Axial computed tomography (CT) scan of the paranasal sinuses showed a clear left facial swelling chronic inflammation and granulomata. Basidiobolus ranarum fungus was isolated on Sabouraud dextrose agar from a biopsy specimen. The organism was characteristic by flat, yellowish-grey, glabrous, becoming radially folded fungus that under the microscope showed broad vegetative hyaline hyphae that bear zygospores with protuberances. The patient made good recovery and was discharged home with no recurrences after receiving oral itraconazole and removal of the polyps surgically.

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MOTS CLÉS
Sinusite ; Champignons ;
Introduction

Fungal species in the order Entomophthorales causes a chronic subcutaneous infection called entomophthoromycosis, which is more common in tropical and subtropical climates [4,6]. Entomophthoromycosis, which includes basidiobolomycosis and conidiobolomycosis, is an uncommon subcutaneous infection with two clinical presentations: subcutaneous zygomycosis and rhinofacial zygomycosis, respectively [6]. Basidiobolomycosis of the nose and face is an uncommon form of zygomycosis in humans. Cases have been noted predominantly in tropical Africa, Asia, or South America [10]. The infection is more common in children, but one case in an adult in India has been reported [2]. It is usually treated with potassium iodide but massive deforming lesions in children can take place. This could be attributed to the lack of early diagnosis and/or improper treatment [5].

This report aims to describe a rare case of rhinofacial sinusitis caused by Basidiobolus species. To our knowledge, this is the first case of rhinofacial mycosis caused by Basidiobolus species in the Kingdom of Saudi Arabia. However, cases have been described in India [1], China [13] and Brazil [10].

Case report

A 22-year-old male patient (hospital #: 1331729/lab. #: as110919) was admitted to Aseer Central Hospital with nasal obstruction accompanied by itching, sneezing, rhinorrhea, epistaxis and recurrent headache. The patient was examined clinically and by computed tomography (CT) scans. Axial CT scan of the paranasal sinuses showed a clear left facial swelling with evidence of sinusitis (Fig. 1). An expansive lesion involving the left maxillary antrum was evident. Involvement with increased density of the left ethmoidal and sphenoidal sinuses has been seen. Right side displacement of the nasal septum with obliteration of the nasal fossa

![Figure 1](image1.jpg)  
**Figure 1** Axial CT scan of the paranasal sinuses of a 22-year-old male patient showing a left facial swelling with sinusitis involving the left maxillary antrum. Right side displacement of the nasal septum with obliteration of the nasal fossa is evident. 

![Figure 2](image2.jpg)  
**Figure 2** Colonies of Basidiobolus ranarum grown on Sabouraud dextrose agar at 30°C for 7 days showing pale grey, wrinkled colonies which are radially and are firmly attached to medium.
also occurred. A soft tissue swelling was noticed on the left temporalis and masseter muscle but adjacent bones were normal.

Cross section biopsy specimen was stained with H&E showed the presence of a chronic inflammation and granulomata. Multiple small soft tissue fragments measuring 1 × 0.5 × 0.2 cm was observed. Mucinous gland hyperplasia covered with respiratory epithelium infiltrated by inflammatory cells (neutrophils, eosinophils, lymphocytes and histiocytes with occasional multinuclear forms) were seen. Few granulomata have been identified. The inflammatory infiltrate encroaches on the vascular walls with wall damage, endothelial swelling and leucocytoclastic fragments of the neutrophil nuclei (nuclear dust). Inflammatory nasal polyp, granulomata and vascular lesions were identified but no malignant cells.

*Basidiobolus ranarum* was isolated from the biopsy material. Colonies become visible after 4 days when cultured on Sabouraud dextrose agar at 30 °C. Colonies were flat, pale grey to creamy-grey, glabrous, becoming radially folded and were firmly attached to the medium (Fig. 2). Microscopic examination demonstrated the presence of broad vegetative hyaline aseptate hyphae, which induced zygosphares which developed protuberances globose one-celled conidia. Two zygophores joined to form sexual zygospores (Fig. 3). The patients received oral itraconazole: 200 mg orally once a day for one month. Polyps were surgically removed during functional endoscopic sinus surgery (FESS). The patient made good recovery and discharged home and follow-up revealed no recurrences.

**Discussion**

It has been reported that fungi are found in 96% of patients with chronic sinusitis and the most commonly isolated fungi are *Aspergillus, Alternaria, Penicillium, Cladosporium, Candida* and *Fusarium* species [9], especially in those who have defective immunities, such as HIV and hepatitis C [3]. During the past 2 decades, zygomycosis become known as a notable fungal infection with distinct clinical patterns and frequently fatal. Within the pathogenic zygomycetes, two orders are of medical and veterinary importance: the Mucorales (*Mucor, Rhizopus, Lichtheimia, and other genera*) which are considered emerging infections in immunocompromised patients with high mortality rates. However, the Entomophthorales (species of *Conidiobolus* and *Basidiobolus*) are rare in immunocompetent hosts and commonly linked with traumatic injuries [7,12].

Entomophthoromycosis in immunocompetent hosts is infrequent and is regularly associated with traumatic injuries. Mortality rates could reach up to 100% subject to patient’s condition. Prompt diagnosis and proper medication of patient underlying medical disorder, surgery, and anti-fungal therapies are prerequisite for a positive result [6]. A combination of oral fluconazole and oral potassium iodide for a total period of 5 months has been applied successfully in a sixty-year-old Indian patient presented with entomophthoromycosis [11].

Basidiobolomycosis is a rare infection; research is needed in southern part of the Kingdom of Saudi Arabia to examine similar chronic rhinofacial and their etiological agents, notably the fungal ones. The present case was treated efficaciously using surgery and antifungal regimens given the positive laboratory feedback. There are many forms of fungal sinusitis and the possibility of a bacterial co-infection was excluded since both the culture revealed pure fungal growth and the treatment was mainly antifungal (itraconazole). Also, the presence of fungus in the sinuses causes an allergic response, resulting in production of allergic mucin and nasal polyps [8], which was evident in the case, presented in this report.

The report highlights the importance of contemplating basidiobolomycosis or zygomyces in general in the differential diagnosis of rhinofacial diseases. Furthermore, we advise that specimens should be routinely submitted from all case of chronic rhinosinusitis to the mycology laboratory.

**Disclosure of interest**

The authors declare that they have no competing interest.
References


