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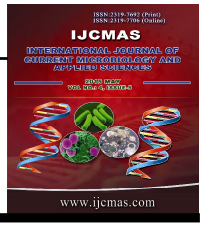

Case Study

Pulmonary Basidiobolomycosis: An unusual presentation in a cancer patient: A case report and mini review of cases in India

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A B S T R A C T

Keywords

Basidiobolo-mycosis, Basidio-bolus ranarum, lung cancer

Basidiobolomycosis is an uncommon disease caused by *Basidiobolus ranarum*. The clinical presentation varies from localized subcutaneous infection to widespread dissemination involving different viscera's, notably the gastrointestinal tract. Pulmonary involvement is rarer; we report a case of pulmonary Basidiobolomycosis in a lung cancer patient.

Introduction

Basidiobolo mycosis is caused by the fungus *Basidiobolus ranarum*, which is a zygomycetes belonging to order Entomophthorales. (Gugnani, H. C *et al*., 1999) This filamentous fungus is usually associated with subcutaneous zygomycosis of trunk and limbs in immune competent individuals (Ribes JA *et al*., 2000) It is an environmental saprophyte isolated mostly from decaying vegetation, foodstuffs, fruits, and soil. It also inhabits the gastrointestinal tracts of reptiles, amphibians, fish, and insectivorous bats. Basidiobolomycosis has been reported worldwide. (Gugnani HC *et al*., 1999). In India most of the reported cases are from Southern part.(Sujatha S et al., 2003; Chandrasekhar HR *et al.,* 1998; Prasad PV *et al.,* 2002; Krishnan *et al.,* 1998) Rare dissemination with visceral involvement by *Basidiobolus* are quoted by various authors such as gastrointestinal tract, uterus, urinary bladder and retro peritoneum. (Bigliazzi C *et al.,* 2004; Khan ZU *et al.,* 2001; Nazir Z *et al.,* 1997; Choonhakarn C *et al.,* 2004) Pulmonary involvement are exceedingly rare, only 3 cases have been reported so far. (Bigliazzi C *et al.,* 2004; Bittenocourt M *et al.,* 198; Ravindran C *et al.,* 2010) We report a case of basidiobolomycosis in an

immunocompromised patient, diagnosed with adenocarcinoma of lung.

Case report

A 59 year old patient diagnosed with adenocarcinoma of lungs few months back, presented to the hospital with chief complaints of productive cough, breathlessness, and fever with evening rise, generalized weakness and inability to walk for two months. He was a chronic alcoholic and smoker. Not a known diabetic or hypertensive and he had no past history of tuberculosis. On examination patient was conscious & oriented afebrile but cachexic, mild pallor was noticed. His blood pressure was 120/80 and pulse rate 96.There was no icterus, clubbing lymphadenopathy or pedal edema. On thorough systemic examination, rhonchii was found on left side of the chest with shift of the mediastinum to the right side. Tenderness was marked over the left hypochondrium. CVS and CNS examination revealed no abnormalities. Laboratory investigation showed Hb 11, TLC-7.9 with eiosinophilia & high ESR (~108). Suspecting tuberculosis sputum was sent ZN staining and AFB culture. Acid fast bacilli were not detected in all the three sputum samples. AFB culture was done using BacTalert, which was reported as no growth later. It was also sent for aerobic bacterial culture and fungal culture, gram stain showed pus cells >25/low power field, Epithelial cells <10/low power field & gram negative bacilli, but no typical respiratory pathogens grew. 10% KOH showed occasional broad hyaline fungal filaments. Chest X-ray PA view showed abnormal opacity in the left upper and parahillar region and fibro nodular lesions were seen in the right midzone suggesting mediastinal mass. CT scan with contrast revealed multiple pulmonary nodules in both lungs with evidence of irregular septal thickening (? Metastasis/? Fungal) & increase in soft tissue densities apart from the malignant mass, measuring about 8.8x 8.3cm arising from the left upper lobe adherent to the mediastinum. There was medistinal shifting to the right with multiple large necrotic lymphnodes in the perivascular, left paratracheal, aorto-pulmonary, left hilar, left tracheobronchial, precarinal, subcarinal region. Left sided pleural effusion (300cc) was also detected. On 4th day filamentous fungi grew in slopes of Sabouraud's and brain heart infusion agars supplemented with chloramphenicol (0.05 mg/ml).Colonies were initially thin, flat, yellowish-grey to creamy grey, waxy, which later became radially folded, reverse was white.[Fig no.1] Lacto phenol cotton blue mount showed broad hyphae with occasional septa and asexual spores. In about 12 days, we could see globose intercalary sexual spores (zygospores) with smooth thick walls and two prominent closely appressed beak-like appendages typical of *Basidiobolus ranarum*.[Fig no.2]

Discussion

Over the last decade or so the clinical profile of medically less important fungal pathogens has taken a widely varied course, ranging from unusual sites, unusual clinical presentations, unusual patient profile, so on and so forth. Basidiobolomycosis is no exception as well. The taxonomy of the '*Basidiobolus* group' has undergone considerable changes, earlier identified as *B. meristosporus* and *B. haptosporus* are now regarded as synonymous with *B. ranarum*.(Gugnani, H. C *et al.,* 1999)

The general disease manifestation involves the subcutaneous tissues of the lower limbs, classically presenting as hardened nodules which expands locally and have eiosinophilic infiltrations.(Gugnani, H. C *et*

al., 1999) Diagnosis relies on histopathology demonstration of the fungal elements with fungal specific stains such as calcuofluor white or GMS and culture. Subcutaneous basidiobolomycosis/ phycomycosis was first described in Indonesia in 1956. (Kian JL *et al.,* 1956) Subcutaneous basidiobolo mycosis has been reported from all parts of India, majority being from South India. (Sujatha S *et al.,*2003; Chandrasekhar HR *et al.,* 1998; Prasad PV *et al.,*2002; Krishnan *et al.,* 1998;. Naniwadekar MR *et al.,* 2009; Rane S *et al.,*;2002; Maiti PK *et al.,* 2004; Ramesh V *et al.,* 2010). However in India, first case was reported by Mukerji *et al.,* 1962 in Bombay (Mumbai). (Thappa DM *et al.,* 2003) Maiti PK *et al.,* 2004detected 7 cases of chronic subcutaneous phycomycosis caused by *B ranarum* during a span of 9 years from 1991 to 1999, from 9 districts in and around Kolkata. Two cases of subcutaneous infections have been reported from western India by Girish M *et al*. 2011 and Sidhi B *et al.* 2007.

Most cases are detected in children and younger individuals. (Table -1) An 11year old girl presented with bilateral nasal block and discharge due to subcutaneous basidiobolomycosis from north eastern India with no particular predisposing factors and a more aggressive course. (Singh R *et al.,* 2008). Mani Anand *et al.,* 2010, emphasized that Entomophthoromycosis should be considered early when children from endemic areas present with unusual, tumor like rapid-growing lesions of the subcutaneous region.

Mendiratta V *et al.,* 2012, reported severe cutaneous zygomycosis due to *Basidiobolus ranarum* in a young infant of 9 months old, who responded to a combination of itraconazole and potassium iodide. Basidiobolomycosis has been reported in as young as 6month old babies following insect bite. (Hema P *et al.,*2013, Anaparthy UR *et al.,* 2014).

No specific risk factors were identified for basidiobolomycosis, mostly reported in previously healthy individuals except in few patients who had a history of diabetes mellitus Ravindran C *et al.,* 2010, Kiran M. Chokka *et al.,* 2010) The Probable mode of infection is trauma or insect bite(Mani Anand *et al.,* 2010; Hema P *et al.,* 2013, Anaparthy UR *et al.,* 2014). Subcutaneous involvement following intramuscular injection has also been described.(Jayanth ST *et al.,* 2013)

However the clinical spectrum of this rare fungus has expanded continuously over past years, a range from chronic subcutaneous lesion to disseminated visceral involvements. Though lower extremity is mostly involved, upper extremity, chest even head, face, paranasal sinuses and neck regions are also involved. (Table 1) Gastrointestinal basidiobolomycosis is a recently described invasive fungal infection, again notably common in pediatric population; clinical findings may mimic malignancy. (Khan ZU *et al.,* 2001; Nemenqani D *et al.,* 2009).

No case reports of GI involvement from India so far, though in one case a 24year old female patient presented with acute intestinal obstruction due to extensive involvement of the soft tissues of the perineum without actual affection of the intestinal mucosa (Angeline N R *et al.,* 2011). *B. ranarum* has been detected as etiological agent of lung abscess in a 42 year old diabetic patient, the only known case of pulmonary origin from India, the precise mode of acquisition of the disease to the viscera remains poorly understood.

Figure.1 Culture of Basidiololus ranarum on Brain heart infusion agar Slant

Figure.2 Zygospores of *Basidiobolus ranarum* that have two closely appressed beak-like appendages

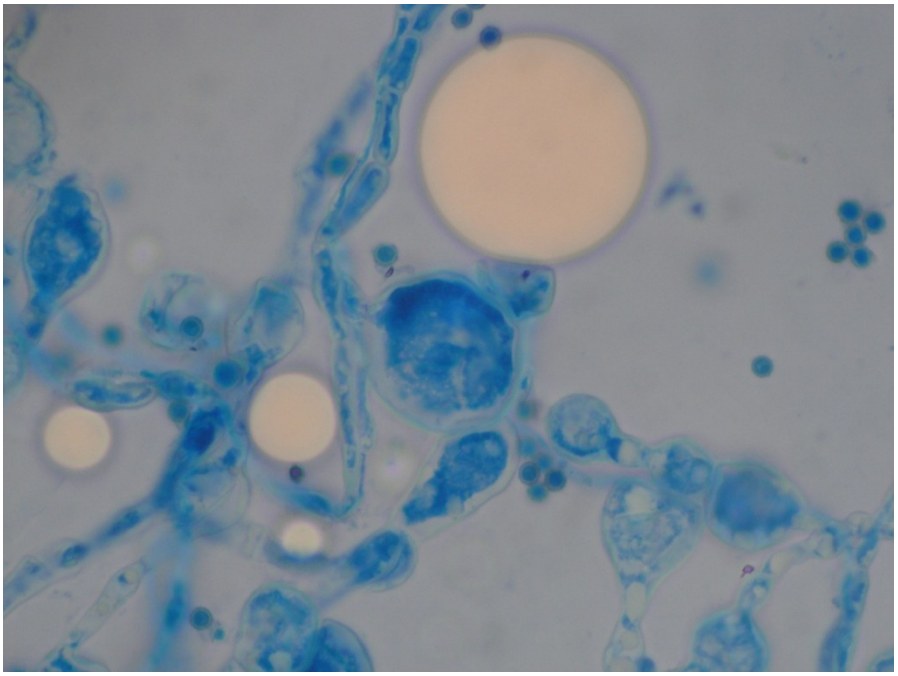
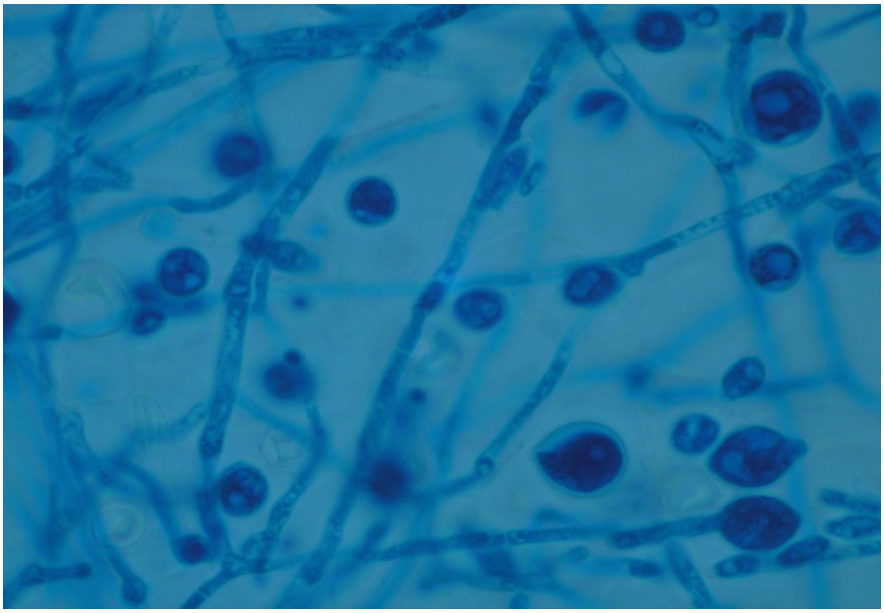


Figure.3 Asexual spores of *B. ranarum*



As per the author the involvement of the trachea bronchial tree could have been caused by gastroesophageal reflux with a primary GI disease (Ravindran C *et al.*, 2010). Disseminated basidiobolo-mycosis was described in an immunocompetent woman, who primarily presented with eiosinophilia and lung infiltrates and subsequently detected to have dissemination on autopsy. (Bigliazzi C *et al.*, 2004) Our patient was an adult with terminal stage cancer, so obviously immunesupressed. In fact the initial clinical differential diagnosis at presentation in our case included neoplasm (especially lung cancer or lymphoma)/ infections especially tuberculosis and allergic bronchopulmonary fungal disease.

During routine evaluation for fever, he was diagnosed to have lung cancer with superadded infection with *B.ranarum*. There was no clue to how he acquired the infection. Leukocytosis, marked eosinophilia, and elevated ESR were found in our case as reported previously in other cases. Some authors have linked pulmonary involvement due to *B haptosporus* as a result of direct extension of the subcutaneous zygomycosis. (Bittenocourt M *et al.*, 1980).

The current experience of treating patients with basidiobolomycosis is limited. The best choice of antifungal agent is not clear, but itraconazole has shown good efficacy. (Thotan SP *et al.*, 2010) Potassium iodide (KI) has been used successfully for treatment of subcutaneous basidiobolomycosis. (Table-1) A combination of both has also been successful in treating patients. (Table-1) However we could not do much for our patient as he succumbed due to cancer.

With the rapid advances in modern medical science, and expansion of fungal disease spectrum we ought to keep in mind the unusual pathogens at unusual sites and the facility for early recognition of such pathogen would definitely help start the right treatment at the right time, saving the life.

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