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A suspected case of chronic Cutaneous Blastomycosis

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So far only three confirmed cases of blastomycosis have been reported from India of which two are culturally confirmed and two are autochthonous cases. So any autochthonous culturally suspected case with classical presentation is worth reporting.

A 67 years old male patient, a resident of Kolkata, who never traveled abroad, presented with multiple warty crusted lesions on both extremities, along with sub-cutaneous nodules on perianal regions and chin for 2 yrs. By KOH wet mount examination, thick wall single budding yeasts with broad base were observed from scraping materials of the margin of lesion. Biopsy tissue cultured in SDCA medium showed fluffy white colonies with a few dumbbell shaped conidia. The patient was non-responsive to various antibiotic therapies but almost cured with 6 weeks of ketokonazole therapy followed by oral potassium iodide therapy for 6 months. However, his facial and perianal lesions recurred after 3 yrs. and were ultimately treated successfully with itraconazole therapy for 9 months.

This is believed to be an autochthonous case of chronic cutaneous blastomycosis, first of its kind reported from India though three other cases with different presentations were reported earlier.

Key words : Blastomycosis, Cutaneous, India, treatment

INTRODUCTION

Blastomycosis is a chronic fungal infection caused by the dimorphic fungus *Blastomyces dermatitidis* characterized by formation of suppurative and granulomatous lesions in any part of the body with a marked predilection for the lungs and skin. The disease has been first described by Gilchrist in 1894 and is therefore also known as *Gilchrist's disease* and Chicago disease. The disease is endemic in North America: Mississippi and Ohio river basins and around Great Lakes. Travelers who visit endemic areas are at a risk of acquiring the disease. Immunocompromised persons are at a greater risk of acquiring the disease (Panakal *et al.,* 2002). Cases have been reported from Africa, Europe, India and Middle East.

Blastomycosis shows a male predominance and is commonest in the thirty to fifty years age group. Pneumonia (rarely detected) is the most common early clinical manifestation, and the lung is almost always the organ initially infected. Cutaneous disease is the most common site of extra-pulmonary involvement and many present with verrucous or ulcerative lesions. Osseous, prostatic, and central nervous system involvements are next most frequent in descending order.

In India, till date approximately twenty-one cases have been reported of which eighteen are not authentic due to inadequate diagnostic evidence. Autochthonous cases of blastomycosis in humans and animals have been mostly reported from Uttar Pradesh (Khan *et al.,* 1982; Randhawa *et al.,* 1983) and Madhya Pradesh (Iyer, 1995; Jambhekar *et al.,* 1988). Two cases of blastomycosis have been reported from Southern India (Ray *et al.,* 1995; Savio *et al.,* 2006). In the present study, a suspected case of blastomycosis with classical dermal manifestation is presented.

MATERIALS AND METHODS

A 67 yrs. old male patient, a resident of Kolkata, with no history of travel abroad, came to the Calcutta School of Tropical Medicine in the year 2000 with multiple warty crusted lesions with serpiginous indurated border containing black dots on heaped up margins and relative central clearing areas on both extremities, perianal region and chin along with sub-cutaneous nodules. The duration of these lesions was two years. He had received various antibiotic therapies during that period with no positive response. On examination if was found that his general condition was apparently good. He was non diabetic and apparently non-compromised, and was not taking any immunosuppressive drugs or steroids.

Routine hematological investigations gave the following results: Haemoglobin was 13 g%, total white blood cell count was 6500/mm^3, platelet count was 19,800/mm^3 and erythocyte sedimentation rate at the end of 1^{st} hrs was 50 mm. His fasting blood sugar was 84 mg/dl, blood urea 21 mg/dl and serum creatinine 1.1 mg/dl. Nothing abnormal was detected in his chest X-Ray. Mantoux test report was negative. Blood reports were negative for HbsAg and HIV and VDRL test result was non reactive.

Scrapings were taken from the edges of the lesions, placed on a clean glass slide and to it 20% KOH was added. After overlying a cover slip, the prepared wet mount was examined under the microscope.

Biopsy material from the lesion was subjected to histopathological study. Periodic acid-Schiff staining, Grocutt's staining and Zeihl Neelson (ZN) staining were done. Unfortunately punch biopsy material was taken by pathologist of a private center from central part of dermal lesion, which was unsuitable for demonstrating fungi present in deep micro-abscess. Biopsy tissue cultured in Sabouraud's dextrose agar with chloramphenicol and incubated at 25°C for four weeks. Mycelial growth was examined microscopically from Lactophenol cotton blue and KOH mounted preparations of the growth. For technical reason yeast conversion test could not be standardized in non-selective Brain-Heart Infusion Blood Agar medium, and same findings were observed for known *Sporothrix schenckii* strains.

The patient was treated with ketoconazole 200 mg table twice daily for a period of six weeks followed by escalating dose of oral saturated potassium iodide therapy up to 20 drops thrice daily for six months. Lesions were almost cured. However, the facial and perianal lesions recurred after 3 yrs. Then he was treated with intraconazole 200 mg tablet twice daily for a period of nine months. Subsequently he was cured completely Thereafter he was followed up every six months and there has been no recurrence after 5 yrs. follow-up.

RESULTS AND DISCUSSION

KOH mounted preparation from the lesions showed thick walled single budding yeasts with broad base (Fig.2) suggestive of *Blastomyces dermatitidis.* Histopathological study showed pseudoepitheliomatous hyperplasia of epidermis with formation of multiple cysts. Adjacent epithelium showed psoriasiform hyperplasia. There was heavy inflammatory cell infiltration adjacent to the epidermis. No malignancy was detected. No mycelia or spores were detected by PAS and Grocott's staining. ZN staining for acid-fast bacilli was negative. Culture of biopsy tissue showed fluffy white colonies in three weeks (Fig.2). Microscopical examination of the growth showed a few dumbbell shaped conidia (Fig.1).

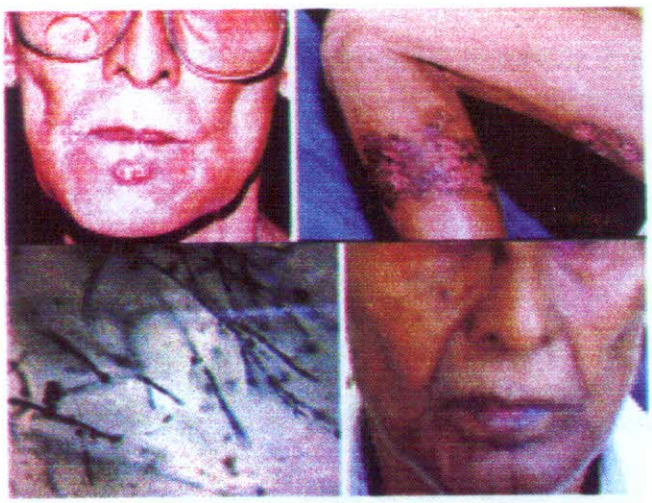

Fig. 1 : (Upper left) Warty lesion of blastomycosis on Chin; (Upper right) Lesions of blastomycosis on lower extremities; (Lower left) Dumbbell shaped conidia of *B. dermatitidis* (LCB mount, 200x); (Lower right) Patient after successful treatment.

Fig. 2 : (Left) Broad based single budding yeast cells in KOH preparation, (50x); (Right) Fluffy white colonies on SDCA.

In India, occurrence of blastomycosis was not known until in 1982 when*B. dermatitidis* was first isolated from the lungs of a bat (Khan *et ul.,* 1982). Thereafter many cases were reported in humans and animals. Of the human cases reported, only three can be taken to be authentic cases. First culture proven autochthonous case was reported in 1983 from bronchial aspirate of a bening pulmonary lesion of a 40-year-old female from Uttar Pradesh (Randhawa *et al.,* 1983). The second authentic case reported in 1988 by Jambhekar *et al.* in a 60 year old female from Madhya Pradesh. She had disseminated proliferative, crusty, ulcerated lesion and was diagnosed by Direct fluorescent antibody test of biopsy material from cutaneous lesion. The third authentic case reported in 2006 in a 41 yr old diabetic male from Bangalore who visited Milwaukee which is an endemic area in the United States of America (Savio *et al.,* 2006). The patient presented with abscesses in left cubital fossa and thigh and had many systemic manifestations. This may not be an autochthonous case.

The case reported by us has the typical clinical picture of cutaneous blastomycosis. The KOH mount was also suggestive of blastomycosis. The clinico-mycological and histopathological findings ruled out sporotrichosis, paracoccidioidomycosis or mycetoma. The laboratory tests also ruled out tuberculosis and tertiary syphilis. The patient had received a number of antibiotic therapies without any positive results but was ultimately cured with ketoconazole and finally itraconazole which is the drug of choice for cutaneous blastomycosis. This is believed to be a autochthonous case of chronic cutaneous blastomycosis, first of its kind reported from India

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