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A Rare Case Report: Primary Cutaneous Blastomycosis with Pulmonary Involvement

Santosh kumar[1], Sweta S. Kumar[2]
PG resident, Department of pulmonary medicine[1], Sarojini Naidu Medical College, Agra[1], Lecturer, Department of Dermatology[2].
Corresponding author: Santosh Kumar, Department of pulmonary medicine, Sarojini Naidu Medical College, Agra.

Abstract: *Blastomycosis is a fungal infection of humans and other animals, caused by theorganism Blastomycesdermatitidis.Blastomycosis has a predilection for involvement of the lungs, skin, bone, and genitourinary tract. We report a unique case of disseminated disease in an immunocompetent male with an unhealed cutaneous lesion on right thigh, who underwent surgical debridement and postoperatively developed life-threatening disseminated blastomycosis progressing to acute respiratory distress syndrome (ARDS). With a high index of clinical suspicion, rapid diagnosis, and prompt therapy with amphotericin B (AmB), the patient recovered.*

Keywords*: Blastomycesdermatitidis, cutaneous lesion, ARDS, amphotericin B.*

I. Introduction

Blastomycosis was first described by Thomas Casper Gilchrist[1] in 1894 and sometimes goes by the eponym Gilchrist's disease.[2] It is also sometimes referred to as Chicago Disease. **Blastomycosis** (also known as "North American blastomycosis", "Blastomycetic dermatitis", and "Gilchrist's disease"[3]) is a fungal infection of humans and other animals, notably dogs and occasionally cats, caused by the organism dimorphic microfungus , Blastomycesdermatitidisa member of the phylum Ascomycota in the family Ajellomycetaceae.Endemic to portions of North America, blastomycosis causes clinical symptoms similar to histoplasmosis.[4,] The disease occurs in several endemic areas the most important of which is in eastern North America, the united States and Canada including the Great Lakes Region and the Mississippi and Ohio River Valleys.[5,6]. Sporadic cases have been reported in continental Africa,[7] the Arabian Peninsula and the Indian subcontinent.Blastomycosis manifests as a primary lung infection.[8]Inhaled conidia can result in a primary lung infection, which may then become disseminated.[9,10].Pulmonary disease is the most common manifestation of blastomycosis with isolated lung disease occurring in 60 to 75 percent of infected people[11] The onset is relatively slow and symptoms are suggestive of pneumonia, often leading to initial treatment with antibacterials. Upper lung lobes are involved somewhat more frequently than lower lobes.[12]. In fewer than 10 percent of cases, blastomycosis can progress to acute respiratory distress syndrome (ARDS) with dyspnea, tachypnea, and hypoxemia as systemic manifestations[13] Palmer and McFadden reported the first case of disseminated blastomycosis causing ARDS in 1968[13].Ifuntreated, many cases progress over a period of months to years to become disseminated blastomycosis and dissemination to skin, bone, genitourinary, and other organ systems can be seen. In these cases, the large Blastomyces yeast cells translocate from the lungs and are trapped in capillary beds elsewhere in the body, where they cause lesions. The skin is the most common organ affected, being the site of lesions in approximately 60% of cases.[14] The cutaneous lesion of blastomycosis is an erythematous indurated area with a chancre associated with lymphangitis and lymphadenopathy. There may be some constitutional reactions with a strong tendency for spontaneous recovery.

II. Case Report

A 44 year old immunocompetent male, farmer by occupation hailing from villlage presented with history of tender, red, solitary subcutaneous nodule over anteromedial side of right thigh (Figure.1). The patient had history of this lesion following minor trauma eight months previously. Over the next three months, he had multiple nodules over the same site (Figure.2) which colasece to form abscess. Right tender inguinal lymphadenopathy present(Figure.3).Testicular examination was unrevealing. As an outpatient, he had been treated empirically with two courses of broad spectrum oral antibiotics. The patient's systemic examination was unsignificant only for subjective fever. He denied chills, shortness of breath, cough, or hemoptysis.

On initial presentation, the patient's temperature was 100.2°F, heart rate 102 beats per minute (bpm), respiratory rate 14, blood pressure 140/70mmHg, and oxygen saturation 97 percent on room air. The chest auscultatory findings were normal bilaterally.Dermatological examinationof the anteromedial right thigh revealed a 8 × 4 cm, soft, tender mass located deep to the subcutaneous tissue.Standard laboratory studies

revealedleukocytosis. Pus culture and sensitivity didn't reveal any bacterial growth. Patient don't respond to the oral antibiotics, then he has given intravenous (IV) vancomycin and ceftriaxone with no relief. The wound was then debrided in the procedure room.

Fivedays after debridement, the patient was noted to be acutely short of breath with a respiratory rate of 24, febrile with a temperature of 103.3°F, and hypoxemic with anoxygen saturation of 74 percent on room air. A chest radiograph revealed diffuse left side pulmonary infiltrates (Figure.4). Because of high suspicion for blastomycosis, another specimen obtained from the cutaneous lesion, was stained with methamine silver, which subsequently revealedthick walled, rounded, broadbased budding yeast (Figure.5). Skin biopsy has taken and sent for histopathology revealedwell-defined granuloma with giant cells contain the round budding cells.(Figure.6). The patient began amphotrecin B (AmB) at a dose of 1mg/kg/day. The patient required supplemental oxygen by face mask, but not ventilatory support. Clinical improvement was gradually seen and the patient was discharged on the 12th day of hospitalization with a six-month course of oral itraconazole therapy (200mg orally twice daily). The patient continued close follow up as an outpatientfor a duration of nine months and is currently fully recovered.

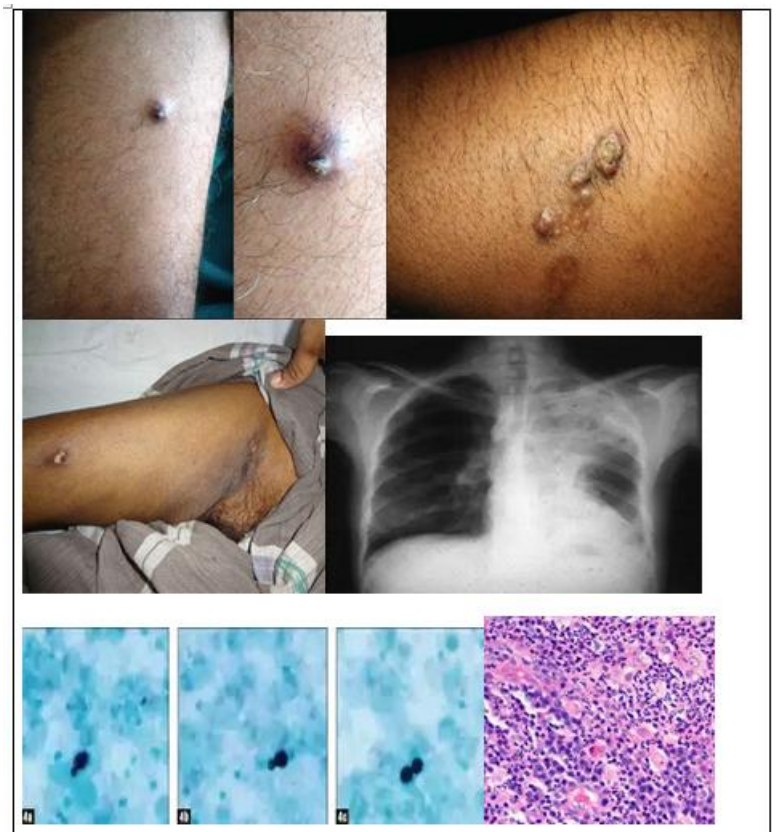

Figure 1.Solitary lesion over thigh (intial presentation). Figure 2.Multiple subcutaneous nodules (after three months).Figure 3. Right inguinal lymphadenopathy. Figure 4. Chest X- Ray showed left side diffuse pulmonary infiltrate. Figure 5. Aspirate from cutaneous lesion stained with methamine sliver stained showed broadbased budding yeast. Figure 6. Histopathology showed well-defined granuloma with giant cells contain the round budding cells.

III. Discussion

B. dermatitidis is a fungal pathogen that can affect any mammalian host. It is a common pulmonary and cutaneous mycosis encountered in people living in North America. The largest cases are reported from Mississippi Valley[15] . few cases reported from the Middle east[15] , India[15] and Poland. In 1951, Schwarz and Baum emphasized that the portal of entry in humans is the respiratory tract rather than the skin, as was previously believed[16] It was discovered that infection occurred by inhalation of aerosolized conidial forms of the organism, which grow in warm, moist soils of wooded areas rich in organic debris.[17], After inhalation, the conidia transform to yeast which, with a thick cell wall, confer resistance to phagocytosis allowing for rapid growth, noncaseating granuloma formation, and progression to an intense inflammatory reaction allowing for dissemination. Extrapulmonary sites of blastomycosis include skin (20–40%), bone (10–25%), prostate and genitourinary organs (5–15%), and the central nervous system (5%). [18]Since this landmark discovery, it has been widely accepted that most cases of cutaneous blastomycosisoccur after hematogenous spread from a

primary pulmonary infection, even in the absence of overt pulmonary disease[19] Most cases of secondary cutaneous infection have no associated pulmonary findings on chest radiograph.[20-22] Primary cutaneous blastomycosis may result as a traumatic inoculation event, but systemic spread fromcutaneous lesions rarely occurs [23]In a patient with cutaneous disease and no evidence of pulmonary disease, current concepts presume an underlying pulmonary infection or one that may have resolved spontaneously. Cellular immunity is considered to be the major protective factor in preventing progressive disease.[24] Skin manifestations of blastomycosis are often very striking, thus the initial cases were reported as primarily dermatologic. Lesions are

more common on the face, neck, and extremities and begin as papules, pustules, or subcutaneous nodules. Typical lesions are verrucous plaques or cutaneous ulcers, frequently with a distinctive purpleblue halo that may suppurate and spontaneously drain, forming deep cutaneous ulcers. The lesions can easily be mistaken for pyoderma gangrenosum, squamous cell carcinoma, and other chronic cutaneous infections, such as sporotrichosis, nocardiosis, atypicalmycobacteriosis, tularemia, anthrax, or leishmaniasis.[25]Pulmonary blastomycosis is less commonly recognized than the cutaneous form.[26] Patients can present with an acute or chronic pneumonia with fever, cough, weight loss, night sweats, and hemoptysis that does not respond to empiric antibiotics. Chest radiographs often reveal diffuse interstitial infiltrates lacking cardiomegaly, pleural effusions, and vascular redistribution; although it is often difficult to distinguish these features from cardiogenic pulmonary edema.[27] In fewer than 10 percent of cases, blastomycosis has a fulminant course manifesting as fevers, chills, and shortness of breath, which can progress to ARDS.Patients often require ventilator assistance within a few days of admission. Many studies highlight the extremely high mortality of ARDS secondary to blastomycosis dissemination.[28]Various antifungal drugs are available for the treatment of blastomycosis. Before antifungal therapy was available, the fatality rate among patients with disseminated blastomycosis was 21 to 78 percent.[29] However, fatality rates dropped significantly after the introduction of AmB in 1956.[30] . Itraconazole is now considered the agent of choice for non-life-threatening blastomycosis with fluconazole, voriconazole, and posaconazole having a role in selected patients.[31] For disseminated blastomycosis, a total dosage of AmB>1g has resulted in cure without relapse in77 to 91 percent of patients, and a total dosage >2g has resulted in cure rates of 97 percent.[32,33]

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