Primary Cutaneous Nocardiosis due to *Nocardia asteroides* Presenting as Mycetoma of Back in an Immunocompetent Patient: A Rare Presentation

Dear Editor,

Nocardiosis is a rare infection caused by gram positive, filamentous aerobic bacteria.[1] It is an opportunistic infection and can present in a localized (cutaneous) or disseminated form.[2] The cutaneous form may either be primary or secondary and can clinically present as a lymphocutaneous form, actinomycetoma, or superficial skin infection.[3] Cutaneous nocardiosis can mimic other cutaneous infections both clinically and morphologically.

A 40-year-old homemaker woman, resident of Uttar Pradesh, India, presented with a painless diffuse swelling, overlying the right side of the back for 4 years. Patient was unable to recall preceding trauma. Her general physical examination was within normal limits. Local examination revealed an indurated, non-tender plaque of size 19 × 31 cm^2 overlying the right back, studded with multiple red to skin-colored nodules and sinuses. There was seropurulent and blood-stained discharge, with intervening areas of puckered scarring [Figure 1a]. Extrusion of granules was not noted following overnight dressings. We considered differential diagnoses of actinomycosis, eumycetoma, actinomycetoma, cutaneous tuberculosis, and an atypical mycobacterial infection. Fine needle aspiration cytology (FNAC) from the lesion showed non-specific inflammatory infiltrate. Deep wedge biopsy on histopathology showed foci of epithelioid cell granulomas with lymphohistiocytic infiltrates. Mycobacterial and fungal cultures did not show any growth. Other investigations (tissue and pus for acid-fast bacilli (AFB), CBNAAT) sent for cutaneous tuberculosis were negative. X-ray and high-resolution computed tomography did not reveal any underlying bone or joint involvement. Based on these investigations, it was difficult to arrive at a conclusive diagnosis, so the patient was empirically initiated on a therapeutic trial of anti-tubercular therapy, with no clinical response at 6 weeks. Histopathology and cultures were repeated and showed fibrocollagenous scar tissue with granuloma, microabscesses, and Splendore-Hoeppli phenomenon in few granules [Figure 2a]. Granules showed PAS positivity with weak acid-fast positivity [Figure 2b]. Repeat culture grew *nocardia asteroides* after 3 weeks of inoculation, seen as acid-fast organisms with beaded branching appearance on AFB stain [Figure 3a] and Gram-positive organisms with branching filamentous appearance on Gram stain [Figure 3b]. Thus, final diagnosis of primary cutaneous nocardiosis (PCN) was made and patient was started on tablet cotrimoxazole (800 mg sulfamethoxazole and 160 mg trimethoprim) twice a day, with marked improvement within 2 months in the form of cessation of discharge and flattening of nodular lesions and complete resolution at 7 months [Figure 1b]. The treatment was further continued for 6 months (a total of 15 months), and the patient is still under follow-up (3 months after stopping treatment) with no recurrence.

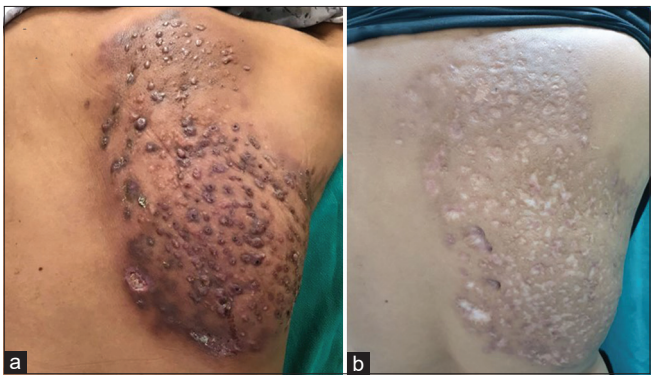

Figure 1: (a) Large plaque over right upper back studded with multiple discrete and coalescent nodules with discharging sinuses (b) Clinical response to treatment after 7 months of treatment

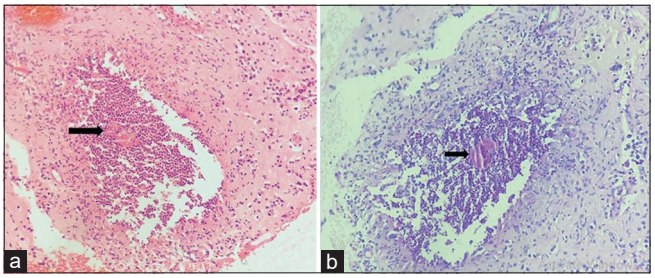

Figure 2: (a) Microabscess showing Splendore-Hoeppli phenomenon (arrow) (H and E, 400x) (b) PAS-positive granules seen in the center of the microabscess (arrow) (PAS,400x)

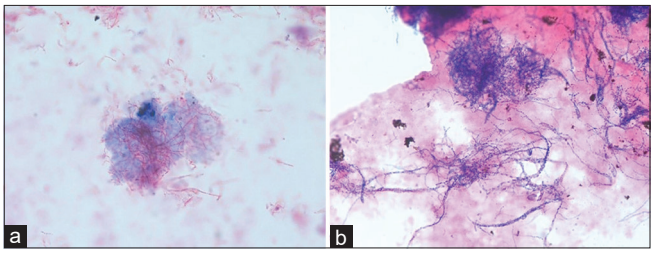

Figure 3: (a) Acid-fast organisms with beaded branching appearance (AFB stain 400 X) (b) Gram positive organisms with branching filamentous appearance (Gram stain, 1000x)

Nocardiosis can be classified into pulmonary (most common), central nervous system, cutaneous, and disseminated forms.[2] Pulmonary and disseminated forms are commonly seen in immunocompromised patients, while the cutaneous form is usually seen in immunocompetent patients.[3] The routes of transmission include inhalation, ingestion, and inoculation.

Cutaneous nocardiosis can be either primary (post-traumatic inoculation) or secondary (as part of disseminated nocardiosis).[4] It has been reported worldwide; however, its exact incidence and prevalence are not known. The most common species causing cutaneous infection is *Nocardia brasiliensis,* followed by *Nocardia asteroides*.[3] Other less common species include *N. otitidis-caviarum*, *N. transvalensis*, *N. farcinica,* and *N. nova*.[2] The risk factors for PCN include accidental trauma, walking bare foot, thorn prick/splinter injury, animal scratch, and immunosuppression.[3]

Clinical variants of PCN include mycetoma (commonest), superficial skin infections, and lymphocutaneous form (rarest). The reported incidence of mycetoma form in India is 5.2-35% with male preponderance.[3,5,6] Extremities are the common sites due to contact with soil and ease of post-traumatic inoculation, with the upper back,[3] scalp, and shoulder being the uncommon sites.[7] The superficial skin infection type has an acute onset with rapid progression and may present as a pustule, an abscess, a bulla, and cellulitis, or as a chronic draining ulcer. The lymphocutaneous variant clinically resembles sporotrichosis.[3] The present case had the mycetoma variant of nocardiosis at an uncommon site (back) with *Nocardia asteroides* as the causative organism. *N.asteroides* as the causative organism in mycetoma form of cutaneous nocardiosis, has not been described in the literature.

Differentials of PCN include cutaneous tuberculosis, actinomycosis, eumycetoma, and sporotrichosis. Acute and inflammatory nature, presence of granules in discharge, small (<1 mm) yellow-white grains, and thin branching filaments on microscopy can help in its differentiation.[8]

A definitive clinical diagnosis of PCN is difficult due to lack of characteristic features. Cultivation of *Nocardia* may take a few weeks due to its slow -growing nature, so it may be reported as negative in the initial few samples.[2] Demonstration of the organism from clinical specimens by Gram stain and modified acid-fast stain are the mainstay of diagnosis. Other serological tests include western blot assay and enzyme linked immunosorbent assay (ELISA), but are of limited use in a resource-poor setting. Radiological investigations in PCN are useful in detecting the involvement of underlying structures.

PCN responds well to medical treatment, and cotrimoxazole is the drug of choice, with clinical improvement seen as early as 3 to 4 weeks; however, complete clearance may take up to a year.[3] Other effective drugs include dapsone, amikacin, amoxicillin, cephalosporins, minocycline, macrolides, fluoroquinolones, and clindamycin.[2] Surgical intervention may be required in patients with multiple discharging sinuses/abscesses.[4]

This case is being reported due to its rare occurrence, chronic extensive lesion at an uncommon site, uncommon organism, and challenges faced in its diagnosis. Since it responds dramatically to treatment, it is imperative for clinicians to have a high index of suspicion in patients with nodulo-ulcerative lesions or discharging sinuses. Moreover, in a tropical country like India, endemic for both tuberculosis and mycetoma, the possibility of cutaneous nocardiosis should always be considered.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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