

CASE REPORT

Cytological Diagnosis of Primary Cutaneous Nocardiosis in a Known Case of Lepromatous Leprosy, Syphilis and HIV

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Abstract:

Nocardiosis is an acute, subacute or chronic bacterial infection caused by a group of aerobic, gram positive and weakly acid fast species of genus Nocardia. Primary Cutaneous Nocardiosis (PCN) is relatively uncommon disease and is usually seen in immunocompetent persons. We report a case of primary cutaneous nocardiosis in a known case of lepromatous leprosy, syphilis and HIV patient who presented with painful swelling over right thigh.

Keywords: FNAC, Immunocomprised host, Nocardia

Introduction:

Nocardia are ubiquitous, gram-positive, filamentous bacilli which are most commonly found in the soil as saprophytes [1]. In humans, Nocardia cause pulmonary, cutaneous, systemic disease and commonly presents as an opportunistic pulmonary disease in immunocompromised person. Infection from the lung can then disseminate to cause systemic disease involving other organs including the skin [2]. Primary Cutaneous Nocardiosis (PCN) is a relatively uncommon disease and is usually seen in immunocompetent persons. PCN occurs either due to wound contamination or deep thorn prick although PCN cases have also been reported without any prior history of injury [3]. The skin lesions include erythematous nodules, pustular ulcers and sinuses. Diagnosis of PCN is challenging due to its varied clinical presentation, inability to detect the microorganisms, slow

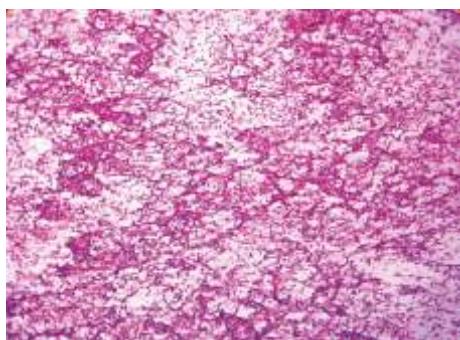
growing nature of the organisms in culture or lack of awareness of PCN [2, 3].

Case Report:

A 35-year-old male, truck driver by occupation, presented with a painful swelling over the medial aspect of right thigh of 15 days duration. There was no history of trauma. The patient was a known case of lepromatous leprosy, secondary syphilis and Retroviral Disease (RVD). Patient was on multidrug therapy for lepromatous leprosy consisting of Dapsone (100 mg daily), Rifampicin (600mg once a month) and Clofazimine (300mg once a month). Patient was receiving Penicillin and Zidovudine for secondary syphilis and for RVD.

On examination, the patient was poorly nourished and febrile. A firm, tender, erythematous swelling was noted over the medial aspect of the right thigh measuring 4x3cm. Fine Needle Aspiration Cytology (FNAC) of the swelling was performed aspirating 1ml of yellowish brown thick pus-like material. Multiple smears were prepared from the aspirate and stained with routine cytological stains along with special stains like Ziehl-Neelsen (ZN) stain. On microscopic examination, cytology smears studied showed cells comprised of sheets of viable and degenerated neutrophils along with few lymphocytes and histiocytes in a background of amorphous necrotic debris and RBCs (Fig.1). ZN stain showed pink filamentous, branching

organisms suspicious of Nocardia. Modified Kinyoun stain showed acid fast branching filamentous bacilli suggestive of Nocardia (Fig. 2). White to cream coloured friable colonies were noted after 48 hours of incubation at 37 degrees on blood agar confirming the Nocardia nova species. Treatment started was cotrimoxazole. Patient responded well and there was no recurrence of lesion with 6 months of follow up.



**Fig. 1: Photomicrograph showing Sheets of Viable and Degenerated Neutrophils in Necrotic and Amorphous Background.
(HandE 100X)**

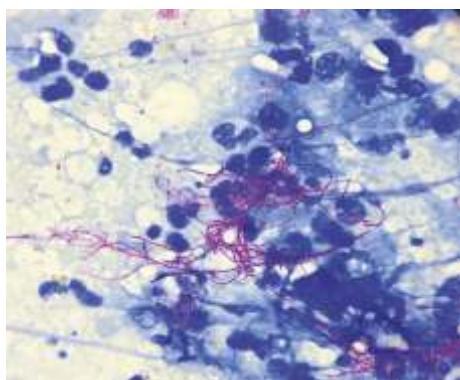


Fig.2: Modified Kinyoun stain showing Acid Fast Branching Filamentous Bacilli Suggestive of Nocardia (1000X)

Discussion:

Nocardia was first described by Edmond Nocard in 1888. Eppinger later described Nocardiosis in humans as a systemic infection [3]. Nocardia belongs to the family Nocardiaceae, order Corynebacterineae and suborder Actinomycetales. Due to their filamentous nature, they can be mistaken for fungi, but fungal hyphae are thicker than the Nocardia filaments. Nocardia are alcohol-acid-fast at some stages of their growth which can further aid in their diagnosis [2, 4].

More than 50 species of Nocardia have been identified, of which 17 species are clinically relevant in humans [2, 5]. *Nocardia brasiliensis* accounts for the majority of the cases, followed by *N. asteroides* which most often causes fulminant systemic infection. Others include *N. otitidis-caviarum*, *N. transvalensis*, *N. farcinica* and *N. nova*. Among these, *N. brasiliensis* can be a fatal complication of HIV and leprosy infection [3].

PCN can occur at any age with cases that have been reported in children as young as three years [3]. It is more common in tropical regions with a slight predilection to the male gender. This can be attributed to the fact that the male population is more exposed to their outside environment than their opposite gender [2]. Nocardia can also occur as an occupational disease in agriculturists and gardeners [6].

The global incidence of nocardiosis ranges from 5% to 24% [2]. An accurate incidence is difficult to determine as it is not an AIDS defined illness, nor is it a notifiable disease [6]. Also, correct identification is difficult as the clinical presentation varies widely creating a diagnostic challenge especially in immunocompromised patients. Culture and Gram stain along with special stains need to be performed to make a diagnosis of Nocardia. Communicating with the

laboratory of a clinical suspicion of Nocardia is important since it is slow-growing and cultures are usually discarded when no growth is seen within 48 hours [2].

PCN occurs as: (1) actino-mycetoma (most common) (2) lymphocutaneous (sporotrichoid) infection, (3) superficial skin infection [3]. Cutaneous dissemination is seen in approximately 20% of patients with systemic nocardial infection. Suspicious skin manifestations of Nocardiosis must include a complete assessment of internal organs as the skin infection might be complicated by dissemination [3]. Primary soft tissue Nocardiosis is a much more uncommon entity than PCN and denotes dissemination of the organisms [7].

Decreased cell mediated immunity is the predisposing factor for Nocardiosis. PCN can also occur as a complication of Crohn's disease. The indolent nature and slow progression may lead to dissemination in other organs and also present with CNS symptoms, such unusual and rare presentations reemphasizes the need to consider nocardiosis as one of the differential diagnosis in an immunocompromised patient who has a history of discharging sinus [8]. Leprosy being a chronic granulomatous infection by itself may have acted as a predisposing factor for concomitant infection with Nocardia in present case. As per Pub Med search till now only two cases of PCN in lepromatous leprosy patient are reported [1-8].

Nocardiosis is a complex diagnosis, because conventional microscopy has its limitations and molecular methods are not routinely available in daily practice. Demonstration of the organism from clinical specimens like granules and pus by Gram stain and modified Kinyoun stain is the mainstay of diagnosis [3]. The most reliable and sensitive method of diagnosis remains tissue culture [2].

Cotrimoxazole for three to four months is the mainstay of therapy for cutaneous Nocardiosis. In systemic form of disease, other effective drugs such as dapsone, amikacin, amoxycillin, cephalosporins, minocycline, erythromycin, ciprofloxacin, imipenem and clindamycin can be used. Mycetoma may require prolonged treatment [1]. Immunocompromised patients are similarly treated, except those who are HIV positive because of adverse cutaneous drug reactions to sulphonamides and thus may need to be treated with alternative drugs [3].

Conclusion:

The incidence of PCN in lepromatous leprosy is very rare and thus presents a diagnostic challenge. This warrants for a detailed clinical examination with thorough sampling and screening of the obtained material by FNAC along with special stains.

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