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Case Report

Coexistent Erythema Nodosum Leprosum with Tubercular lymphadenitis in an Immunocompetent Individual: An Unusual Association

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Abstract

Leprosy and tuberculosis are chronic granulomatous diseases known to be endemic in our country. Simultaneous occurrence of both infections in a single individual is not an uncommon clinical condition but has been reported infrequently in literature. Erythema Nodosum Leprosum (ENL) is an immunologically mediated hypersensitivity reaction presenting in patients with lepromatous and borderline lepromatous leprosy usually before, during and rarely after multidrug therapy (MDT) for leprosy. Here we report a case of ENL and tubercular lymphadenitis diagnosed simultaneously in a 25 year old young male presenting with fever and other systemic features at the time of admission in our hospital. It is important to identify these cases to avoid monotherapy of tuberculosis. Concurrent occurrence of these diseases can modify the course and presentation of each other and pose diagnostic and therapeutic challenges. Due to relative rarity of occurence of this phenomena we are reporting this case.

Keywords: Erythema Nodosum Leprosum; Tubercular Lymphadenitis.

Introduction

Both leprosy and tuberculosis are chronic granulomatous diseases known to be endemic in India. The occurrence of both diseases concurrently in a single individual is not an uncommon clinical condition but has been reported infrequently in literature [1]. The infrequent occurrence of both tuberculosis and leprosy together is based on the transmission dynamics of these infectious diseases [2].

Erythema nodosum leprosum (ENL) or type 2 reactions are usually serious, often difficult to manage, immune-mediated complications of lepromatous leprosy (LL) and borderline lepromatous (BL) leprosy [3]. It presents in patients

usually before, during and rarely after multidrug therapy (MDT) for leprosy. Patients with ENL present with fever, malaise, and crops of painful erythematous nodules [4]. Many other systems are affected in ENL resulting in iritis, neuritis, myositis, lymphadenitis, arthritis, dactylitis, and orchitis [3]. The underlying pathology is of antigen-antibody immune complex deposition.

Here we report a case of ENL and tubercular lymphadenitis diagnosed simultaneously in an individual at the time of admission in our hospital. To the best of our knowledge, this is the first case report of coexistence of ENL in tuberculous patients. It is important to identify these cases to avoid monotherapy of tuberculosis.[1] Due to relative rarity of occurence of this phenomena we are reporting this case.

Case Report

A 25-year-old man was admitted with complaint of fever off and on since 2 years. It was associated with joint pain, rash over face and dysuria since seven days. In the past history the patient was worked up for tuberculosis at a private clinic and Antituberculous treatment was started which he was taking since past 5 months. He was not very compliant with drug intake.There was history of epistaxis, watering of eye and ocular irritation present.

On examination he was thin built weighing 46 kg. He had bilateral axillary and left cervical lymphadenopathy with erythematous plaque over nose, bilateral cheek and forehead. Pain and touch sensation was intact over these lesions. Few tender nodular eruptions of varying size were present over trunk and upper extremities. The scalp and oral mucosa were not involved. Bilateral ulnar, radial and common peroneal nerve, left supraorbital and right posterior tibial nerve were enlarged and tender. He also had episcleritis and migratory polyarthritis. At the time of admission his temperature was 40°C, blood pressure 120/70 mm of Hg, and pulse rate 114/min and respiratory rate was 38/min. Respiratory system and rest of the systemic examination was normal.

Investigations revealed Hb 12.9 gm%, total leucocyte count 8600/mm^3 (P 85%, L 15%), ESR 26 mm in the first hour, platelet count 2.2 lakhs/mm^3, renal and liver function tests within normal limits. His S. calcium was 8.2 mg/dl, S. Sodium 131 meq/L, S.potassium 4 meq/l. Peripheral smear showed normocytic normochromic cells with no evidence of hemolysis. Anti Nuclear antibodies was negative, Angiotensin converting enzyme was increased. Anti ds DNA Abs was 0.16. Toxoplasma IgG was 0.25.

Slit-skin smear from the erythematous nodules revealed clumps of Acid Fast Bacilli on Fite stain (Bacillary Index-5) and thus tested positive for lepra bacilli. X-ray chest was normal and HIV status was negative. Fine Needle Aspiration Cytology (FNAC) of axillary lymph node was done which revealed numerous epithelioid granulomas in a necrotic and inflammatory background. Ziehl Neelsen stain showed many Acid fast bacilli (Fig. 1). Thus based on the above findings a diagnosis of Tubercular lymphadenitis was given. Gene expert was done for mycobacterium tuberculosis which was positive. Skin biopsy from right forearm showed collection and sheets of foamy macrophages in dermis. Mixed dermal infiltrate of neutrophils and lymphocytes was seen superimposed on foamy macrophages. Many dispersed epithelioid cells were seen with foci of necrosis and multinucleated giant cells (Fig. 2). Special stain for lepra bacilli ie. Fite-Faraco stain was positive.

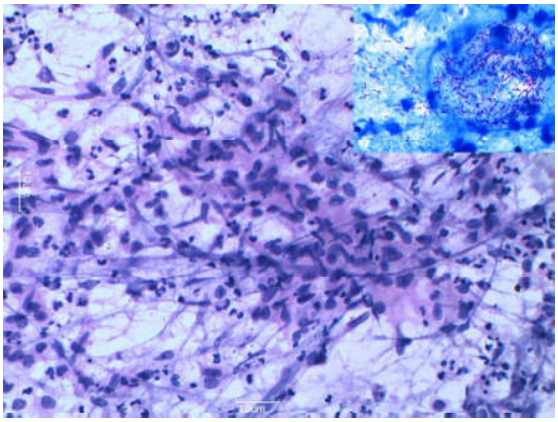

Fig. 1: Smear shows epithelioid granuloma lying in a necrotic and inflammatory background.(MGG stain, 40x). Inset shows numerous acid fast bacilli.(ZN Stain, 100x)

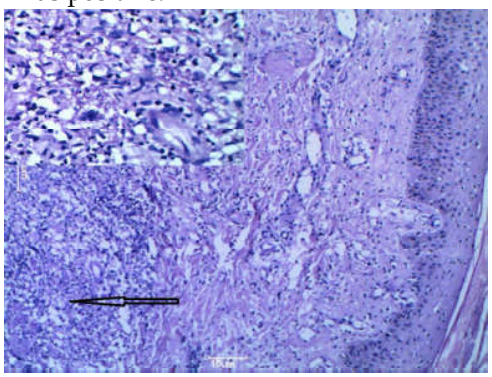

Fig. 2: Section shows sheets of foamy histiocytes lying in the dermis (arrow) and separated from epidermis by clear zone. Inset shows mixed dermal infiltrate of neutrophils and lymphocytes along with scattered epithelioid cells and a multinucleated giant cell. (H&E stain, 40x)

With the clinical scenario and investigation results, final diagnosis of Erythema Nodosum Leprosum with tubercular lymphadenitis was made. Multidrug therapy was started along with steroids for ENL Patient was also put on anti tubercular treatment. General condition of the patient improved. Patient was discharged on fifth day and is on regular follow up.

Discussion

Erythema nodosum leprosum (ENL) or type 2 lepra reactions complicate lepromatous leprosy and borderline lepromatous leprosy. The overall prevalence of ENL was 24%; 49.4% among cases of lepromatous leprosy (LL) and 9% among cases of borderline lepromatous (BL) leprosy [3]. Most of the cases have ENL after having started multidrug therapy. However, some patients have *de novo* ENL without any apparent previous treatment as seen in the index case.

The concomitant occurrence of both tuberculosis and leprosy in a single individual is not an uncommon clinical condition but is being reported infrequently in literature. The infrequent occurrence of both tuberculosis and leprosy is based on the transmission dynamics of both infections. The higher reproductive rate of *Tubercle bacilli* as compared to *Lepra bacilli* and degree of cross immunity within an individual prevents the simultaneous occurence of both the infections [2]. Chaussinand *et al*. concluded that the prevalence of leprosy was inversely related to the prevalence of tuberculosis. There have been few sporadic reports of coexistence of tuberculosis and leprosy in the same patients. In 1895, Hansen observed that tuberculosis was a major cause of death in his leprosy patients. Kumar *et al*. reported that tuberculosis may occur through the spectrum of leprosy [4]. In leprosy, majority of the cases reported were of pulmonary tuberculosis and very rarely extra-pulmonary tuberculosis has been reported [5]. In our case, patient did not show any pulmonary involvement.

However, the coexistence of Erythema Nodosum Leprosum with tuberculosis has been very rarely reported. We could find only two case reports in the literature where Erythema Nodosum Leprosum (ENL) was detected in a Tuberculosis patient. While one case was associated with Pulmonary Tuberculosis [1], the other was associated with Perianal Tuberculosis [6]. We could not identify a report where ENL was associated with Tuberculous Lymphadenitis. It is believed that the reduction in an effective cell-mediated immune response associated with multibacillary leprosy lead to reactivation of an underlying latent tuberculosis infection, or to superinfection with *M. tuberculosis* [2].

Type 2 lepra reaction is an inflammatory systemic reaction having varied presentations in which immune complexes can deposit in any organ or tissue [7]. Neuritis, orchitis, uveitis, periostitis, lymphadenitis, arthritis and glomerulonephritis can occur rarely. There have been reports of increased incidence of ENL in Lepromatous patients if injected with intradermal IFN gamma or PPD [8,9]. It is well documented that ENL is associated with multidrug therapy and treated with corticosteroids. So there is a documented immunological basis of ENL in Leprosy. In our case patient developed painful erthematous nodules and also showed episcleritis and arthritis.

Approximately one-third of leprosy patients may present with Type II lepra reaction (erythema nodosum leprosum) at the time of diagnosis. In leprosy lymph node involvement can be due to Type II reaction or as a part of visceral involvement in lepromatous leprosy [10]. Coexistent lepromatous and tuberculosis infections involving lymph nodes have been reported apart from tubercular lymph node involvement in patients of leprosy [11].

Most of the cases of tuberculosis are associated with lepromatous leprosy followed by borderline lepromatous leprosy, as observed in present case also. However, few authors have reported the association of tuberculoid type of leprosy with tuberculosis [12]. Only two cases of tuberculosis were reported to occur earlier than leprosy [13]. One study concluded that tuberculosis can occur during full spectrum of leprosy [14]. It is important to identify the coexistence of tuberculosis with ENL to avoid monotherapy in these cases as along with the multidrug theapy (rifampicin, clofazimine and dapsone), antitubercular treatment also needs to be instituted to achieve complete cure.

To conclude, we would like to emphasize upon the importance of diagnosing coexistent leprosy and tubercular lymphadenitis in an endemic country like India. Concurrent occurrence of leprosy and tuberculosis can modify the course and presentation of each other and pose diagnostic and therapeutic challenges. Furthermore, coexistence of ENL with Tuberculosis should lead to further research on the cause of this association and how to go about multi drug therapy.

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