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**Case Report**

# Father-Daughter Duo: How Unmasking Leprosy in Father Helped in Leprosy Diagnosis in the Daughter - A Case Report

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In highly endemic countries like India, where tuberculosis (TB) and leprosy infection may coexist, screening the other disease before initiating treatment is important to prevent Rifampicin resistance since both diseases are treated with and sensitive to Rifampicin. Here, we report a leprosy case involving the unmasking of leprosy in a treated patient with Pulmonary TB. In this case, a high index of suspicion of Erythema Nodosum Leprosum (ENL) in a patient with no history of leprosy disease or treatment with anti-leprosy drugs was observed. He, however, had a history of taking anti-tuberculous medicine 1.5 years earlier. This case report also acknowledges the physician's prompt referral of this patient to a dermatologist. Taking a detailed family history and screening helped us diagnose leprosy in the patient's daughter. It also emphasises the atypical presentation of leprosy, which (although described in textbooks) is being reported here.

**Keyword**s: Hansen's disease, Tuberculosis, Erythema Nodosum Leprosum, Contact Tracing, Rifampicin

## Introduction

Tuberculosis (TB) and leprosy infection may occur together in the same patient in highly endemic countries (Kama et al 2019). Both leprosy and TB are prevalent in India and are sensitive to treatment with Rifampicin (RMP). TB must be ruled out in cases of leprosy before treatment is initiated to prevent RMP resistance in TB (Mangum et al 2018).

Type 2 lepra reaction/ ENL (Erythema Nodosum Leprosum) is an immune complex syndrome causing inflammation and deposition of immune complexes in the skin (presenting as nodular swellings with inflammation), nerves (neuritis) and other organs, along with other constitutional symptoms like fever, and lymphadenopathy. It is usually observed in multibacillary patients, before treatment (as is in this case), during therapy with anti-leprosy drugs and even after release from treatment (Nath 2017).

Here we discuss a leprosy case involving the unmasking of leprosy infection by diagnosing ENL in a *de novo* case, which led us to diagnose leprosy in his daughter too.

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## Case Report

A 40 years old male was referred to Dermatology Out Patient Department (OPD) for acute eruption of skin lesions associated with fever for three days. The lesions were noted first on the arms first and quickly encompassed the chest, abdomen and lower limbs, along with fever. The patient consulted a physician for his symptoms and was treated for fever and joint pain. He was referred to a dermatologist for his skin lesions without any clue as to the probable diagnosis. Clinical differentials of erythema nodosum, likely to be drug-induced/ leprosy/ tuberculosis, were considered.

There was no history of any recent drug ingestion or history suggestive of drug hypersensitivity. There were no complaints of accidental slippage of footwear, difficulty in buttoning-unbuttoning the shirt or tying shoe laces (no history suggestive of reduced grasping power), accidental or unidentified trauma or burns.

On examination, no hypopigmented patch was identifiable. An ichthyotic patch was present on the right knee. Multiple erythematous, tender, nodular lesions were present on the upper half of both the extremities (Figure 1a) and trunk (Figure 1b). The palms, soles, scalp, and mucosae were spared. The patient was evaluated for evidence of leprosy; cutaneous examination showed infiltration of bilateral ear lobes and eyebrows (Figure 2) and lateral superciliary madarosis. His peripheral cutaneous nerves, including supraclavicular, ulnar, radial cutaneous, lateral popliteal, and posterior tibial nerves, were thickened and non-tender with glove-stocking anaesthesia. He did not have any signs suggestive of neuritis. Mild motor impairment for small muscles of the hands was present. There was no evidence of deformity or non-healing trophic ulcer. No past history of treatment with Anti-Leprosy Drugs (ALD) could be obtained.

On exploring the other differential diagnoses, active TB was ruled out by clinical examination and chest X-ray. On probing further, this patient was a treated case of TB, having taken AKT for six months before 1.5 years. He also has diabetes mellitus (DM), diagnosed 2.5 years earlier, under control with treatment.

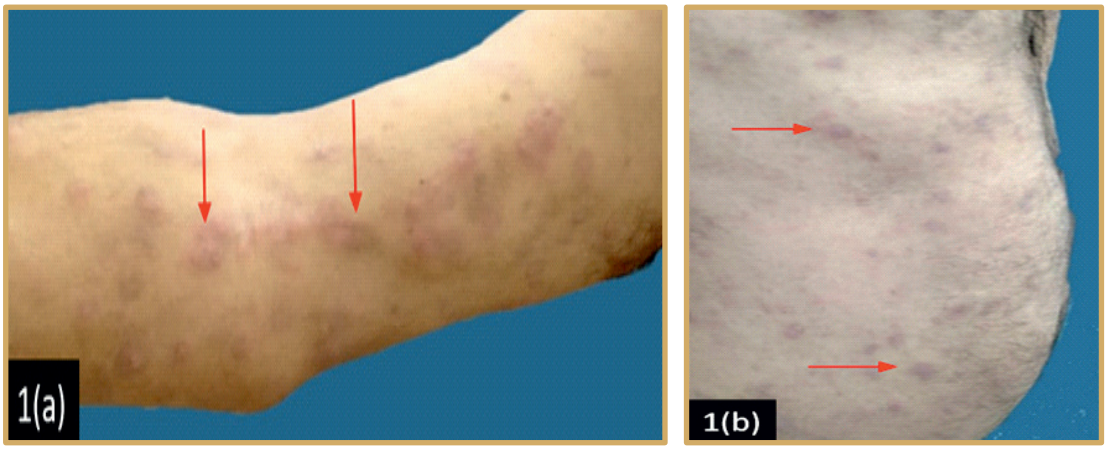


**Figure 1a & 1b : Clinical photograph showing multiple erythematous nodules (ENL) on the upper extremities & trunk in a patient of leprosy with past history of Anti-Tuberculous treatment.**

After informed written consent, a full-thickness skin biopsy (from an erythematous nodule over the right lower back) was taken. Considering the probable diagnosis of ENL reaction with MB leprosy, a slit skin smear (SSS) from both the earlobes and eyebrows was also obtained. The Ziehl–Nielson stained SSS was negative for AFB (acid-fast bacilli). Still, skin biopsy was suggestive of ENL due to leprosy, with a perivascular and peri-adnexal collection of lymphocytes and macrophages (with foamy cytoplasm) in the dermis (Figure 3a). It also showed the presence of sparse solid-staining and multiple fragmented bacilli (single and in bundles) on the Wade-Fite-Faraco stain, with a bacillary index (BI) +4 (Figure 3b).

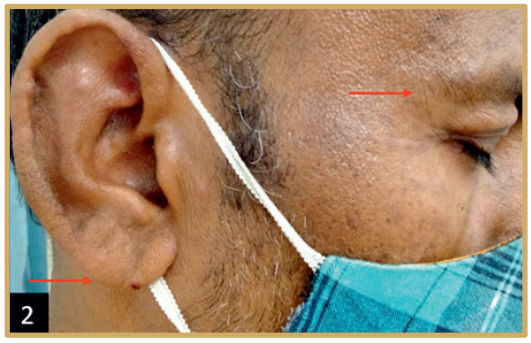


**Figure 2 : Ear lobe infiltration and superciliary madarosis in case of leprosy with Erythema Nodosum Leprosum (ENL)**

The patient showed no active lesions suggestive of leprosy but showed ENLs, and was diagnosed with lepromatous leprosy. The organ involvement of hepatitis, glomerulonephritis or epididymal-orchitis was ruled out; the patient was prescribed WHO-recommended multidrug therapy (MDT) for multi-bacillary (MB) leprosy. Additionally, oral prednisolone was added to his regimen to treat the type 2 lepra reaction. The patient has been on regular follow-ups once a month and has recently completed his treatment of 12 MB MDT

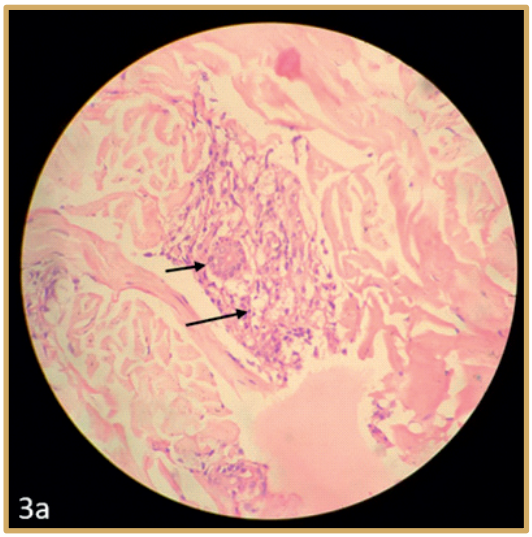


**Figure 3a : Figure showing periadnexal infiltration of foamy macrophages and lymphocytes in dermis in the biopsy taken from the patient (H & E stain, 40 X magnification).**

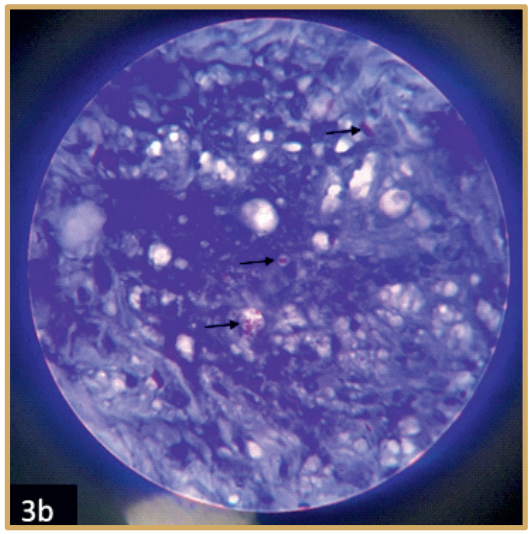


**Figure 3b : Figure showing fragmented acid-fast bacilli on Wade-Fite-Faraco stain, bacillary index 4+ in the same biopsy sample of the patient (Fite stain, 100 X magnification).**

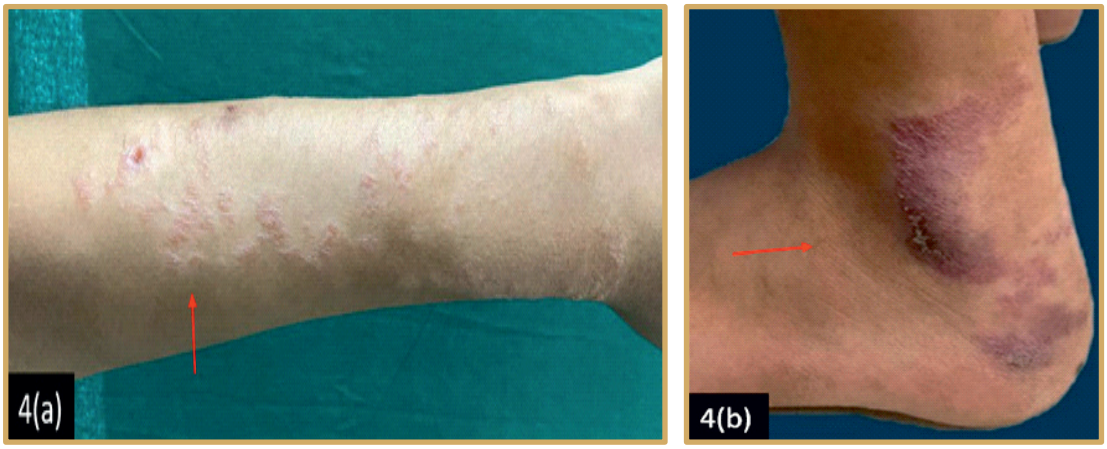


**Figure 4a & 4b : Erythematous annular patch with satellite lesions on left forearm & left foot in daughter of the patient (father) having de novo Erythema Nodosum Leprosum (ENL).**

blister packs. After the initial management of ENL lesions with oral corticosteroids, there was no recurrence of similar lesions.

On screening the family members, the 19-year daughter of this patient was found to have an erythematous patch with satellite lesions on both left forearm (>10 cm in length) and left foot (7-8 cm long) for 1.5 years. On careful examination, two hypopigmented patches with elevated erythematous scaly margins, characterised by satellite lesions around the patch, were present. Considering two patches with satellite lesions and elevated margins, the patient was put into the borderline category. On neurological examination, the ulnar nerve on the ipsilateral (left) side was thickened, cord-like and tender. Neuritis was present in the left ulnar nerve. Examination of other nerves was non-significant. Looking at the size of the lesion (>10 cm), the patient was prescribed MB MDT (Figures 4a & 4b). Absence of sensations over the lesions, neuritis and positive family history of leprosy (in the father) supported the diagnosis. The slit skin smear was negative, but a biopsy revealed lymphocytic infiltration, perineural and periadnexal granuloma formation, epithelioid cell collection in the dermis (negative Wade-Fite-Faraco stain), confirming the diagnosis of Borderline leprosy. The daughter is currently under treatment and on regular follow-ups and will be released from therapy (RFT) soon.

The family consisted of three family members, of which the father and daughter were diagnosed with leprosy and treated/ undergoing treatment. As a part of contact tracing, the mother was screened for leprosy with no clinical findings suggesting the same.

**Discussion**

Though leprosy and TB are believed to co-occur in highly endemic countries like India, their coinfection is not frequently encountered in clinical practice (Shetty et al 2018).

Leprosy and TB and their repercussions on the incidence of each other remain a matter of debate even in endemic countries (Trindade et al 2013). The immunological milieu of the host appears to paradoxically influence susceptibility to mycobacterial coinfection with no consensus

regarding whether prior exposure to one offers protection or predisposition to the other (Mangum et al 2018).

The clinical implications of failure to identify coinfection cannot be understated (Mangum et al 2018); this unusual case reflects the same.

The dosage, as well as the duration of treatment with Rifampicin, varies in TB and leprosy. Therefore, before beginning leprosy treatment, we must screen for active TB disease. Although few specialists and Institutions undertake this, vice versa, i.e. screening for leprosy is not done before starting anti-tuberculosis treatment in patients diagnosed with TB.

Clinicians need to be aware that leprosy and TB may occur concurrently, and they may need to rule out one if the other is present. A thorough history and examination, and good clinicopathological correlation are necessary for diagnosing atypical dermatoses or common dermatoses with unusual presentations. Instituting once-a-month RMP treatment in an undiagnosed TB coinfection in a leprosy patient has the risk of inadequate therapy, which may result in RMP resistance. This has also been emphasised and highlighted, as this subsequently increases the chances of developing resistant mycobacterial strains (Masuka et al 2021).

The history of pulmonary tuberculosis treated with anti-TB drugs could be responsible for this unusual presentation of the patient with ENL lesions, with no history of having taken ALD. Rifampicin was the only drug (no MDT), given for the first two months of anti-TB treatment, and no effort was made to look for signs and symptoms of leprosy.

ENL may lead to neuritis, leading to deformities and gross morbidities, with a massive impact on the quality of life. Additionally, ENL being a multisystem disorder, a patient can present with diverse systemic manifestations. Hence it becomes imperative for practitioners of all specialities to keep in mind the probability of the same and should be considered. Timely diagnosis of this unusual leprosy case helped pick up another case, which was otherwise misdiagnosed and deprived from ALD; this time – the teenage daughter of the patient.

Family screening is an integral part of leprosy management which helps in early detection and treatment. Examining the household contacts includes not only the family members but should extend to the neighbours and other people in the neighbourhood, wherever possible. The risk of developing leprosy is five to ten times in those with a family member with the disease. The risk is higher in lepromatous leprosy and lower in tuberculoid leprosy. The high rate of leprosy in household contacts emphasises the continuing need to screen and follow up on the asymptomatic contacts of newly diagnosed leprosy patients (Ramasamy et al 2018). Follow-up of contacts is an efficient method of case detection of leprosy in the general population (Ramasamy et al 2018). The literature suggests that targeted interventions should be aimed at close contacts (Moet et al 2004). As per NLEP 2020 guidelines, the close contacts of every index case of leprosy shall be screened for signs and symptoms of leprosy, and these contacts shall be administered with a Single Dose of Rifampicin (SDR) as post-exposure Chemoprophylaxis (PEP) (NLEP 2020).

## Conclusion

Clinical suspicion of ENLs in any setting needs further evaluation to unmask the underlying disease, which in this case was leprosy. Hence, a timely dermatology/skin clinic referral rules out leprosy and prevents further morbidity leading to a better prognosis for the patient. Also, identifying leprosy-TB coinfection is crucial for managing this dual infection as it is for the future considerations

of drug resistance to a highly bactericidal agent Rifampicin.

Household contact screening is an effective method for case detection in leprosy elimination. Identifying the contacts of leprosy patients at high risk of the disease is of utmost importance to break the transmission of the disease, as well as early diagnosis and treatment of leprosy patients.

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