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Case Report

Lateral Cutaneous Nerve of Forearm: A Rare Cutaneous Nerve to Develop Abscess in a Relapsed Case of Leprosy

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Leprosy is a disease of the skin and nerves; and the involvement of both peripheral and cutaneous nerves is often found in leprosy. Despite adequate treatment with Multi Drug Therapy, Mycobacterium leprae may survive and cause relapse, which is characterized by a combination of new signs or symptoms and presence of acid-fast bacilli on skin or nerve biopsy samples. Nerve abscess is an unusual presentation of leprosy relapse, and has been reported most commonly in ulnar nerve followed by median nerve and common peroneal nerve and rarely in cutaneous nerves or nerves supplying a patch of leprosy. We report a rare case of nerve abscess in lateral cutaneous nerve of forearm in relation to a leprosy patch, in a relapsed case of Borderline tuberculoid leprosy.

Keywords : Nerve Abscess, Lateral Cutaneous Nerve of Forearm, Relapse, Leprosy

Introduction

Leprosy is characterized by involvement of nerves by primary infection and by immunologically mediated reversal reactions. Nerves involved in leprosy can either be major nerve trunks and their branches or cutaneous nerves. The presence of abscesses in the involved nerve depends on the type of immunological reaction of the host. Abscesses are more common in Paucibacillary (Tuberculoid and Borderline Tuberculoid) leprosy and commonly involved nerves are the greater auricular, radial, ulnar, median, and common peroneal nerves (Kumar et al 1997). Cutaneous nerves of forearm include lateral, posterior and medial cutaneous nerves of forearm innervating skin of forearm. These thickened nerves are generally innervating anaesthetic skin lesions and likely to be missed unless palpated for carefully. Nerve abscesses have rarely been reported in these cutaneous nerves. However, in cases of tuberculoid spectrum of leprosy or reversal reactions, nerve abscesses can develop in these nerves due to exaggerated immunological reactions (Char & Cross 1986).

Treatment of leprosy is unique in terms of fixed dose and duration of regimens and often,

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termination of treatment is based on completion of the recommended duration of treatment rather than disappearance of clinical signs and symptoms leading to relapse (Kaimal & Thappa 2009). We report a case of nerve abscess in lateral cutaneous nerve of forearm in a case of borderline tuberculoid leprosy, previously treated with Multi drug therapy (MDT) containing Rifampicin and Dapsone for six months, and subsequently developing relapse of leprosy patch and nerve abscesses one year after release from treatment. This case also highlights the inadequacy of uniform six-month two drug regimen for all paucibacillary (PB) leprosy cases and the need for continuous monitoring of disease progression and activity before release from treatment.

Case Report

A 28-year - old male, an armed forces soldier, presented with complaints of light-colored numb patch over left forearm of two years duration for which he had previously been treated with two drug MDT for 6 months containing Dapsone and Rifampicin with partial resolution of skin patch post treatment. However, one year after release from treatment, patient reported at our center with complaints of increase in size of numb patch with painful thickened cord liked lesion around the patch which were insidious in onset and gradually progressive. Examination at presentation revealed a solitary, well defined, hypopigmented, hypotrichic, hypohydrotic patch over left forearm, with streaming borders, measuring 15 cmx 10 cm which was hypo-aesthetic to pain touch and temperature (Fig 1). Thickened tender nerve to patch was palpable along the periphery of patch on the superior aspect. Left ulnar nerve was grade 2 thickened and non-tender. No other lesions were present elsewhere on body. Slit-skin smear from the right ear lobe as well as lesion, stained using Ziehl–Neelsen (ZN) stain was negative for acid fast bacilli (AFB). An elliptical

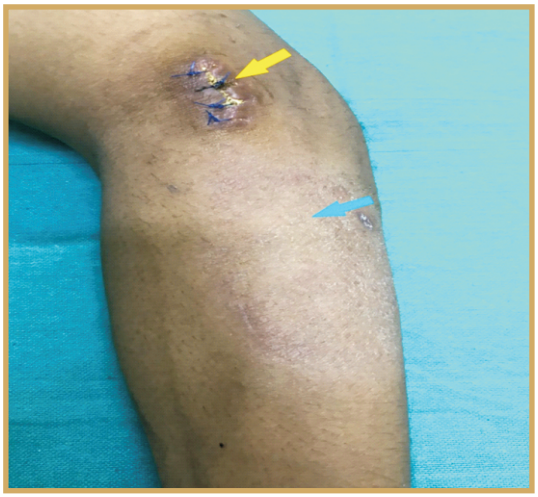

Fig. 1 : Well defined hypopigmented, hypotrichic, hypohydrotic patch over left forearm with streaming borders measuring 15 cm x 10 cm (blue arrow). Thickened nerve to patch (post nerve decompression and biopsy) (yellow arrow).

incisional skin biopsy was done from the lesion over left forearm which revealed the following. Epidermis showed occasional flattening of rete ridges. Dermis showed infiltration by lymphocytes, with many well-formed granulomas comprising histiocytes and lymphocytes. Minimal appendages and nerve bundles were seen. Granulomas were seen around dermal appendages and neurovascular bundles (Figs. 2a and b). No AFB was demonstrated in the tissue section using modified Ziehl-Neelsen stain. An ultrasound followed by MRI of affected area was done to delineate affected thickened cutaneous nerve around the patch which revealed a well-defined ovoid branching focus of altered signal intensity continuous with the nerve in the subcutaneous tissue on lateral aspect of distal third of arm measuring 66.6 X 22.3 X 8.3 mm in size (Figs. 3a and b). On the basis of clinical and histological

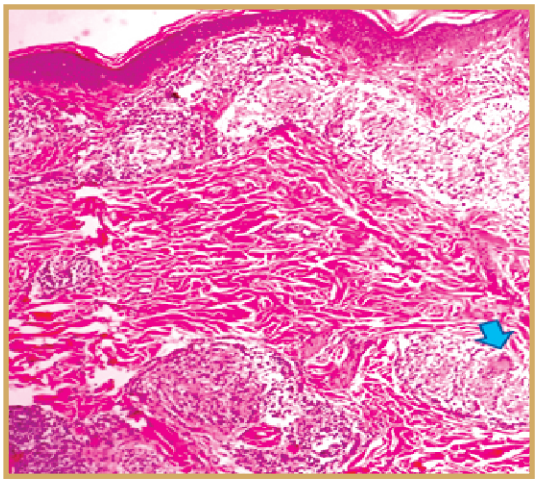

Fig. 2a : Skin biopsy : Hematoxylin & Eosin stain (40X): Infiltration of dermis with many well formed granulomas (blue arrow) comprising histiocytes and lymphocytes around dermal appendages and neurovascular bundles.

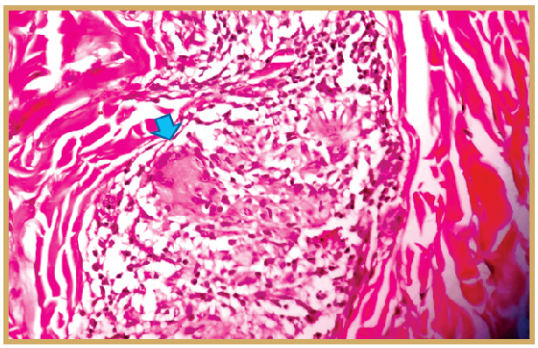

Fig. 2b : Magnified view of granuloma with langhans giant cells (blue arrow).

features patient was diagnosed as a case Borderline Tuberculoid leprosy-Relapse, and was restarted on 3 drug MDT along with oral steroids in view of tender nerve abscess. However, due to persistent pain, one-week post initiation of steroids, patient was taken up for nerve de-compression. Intra operatively, the abscess was

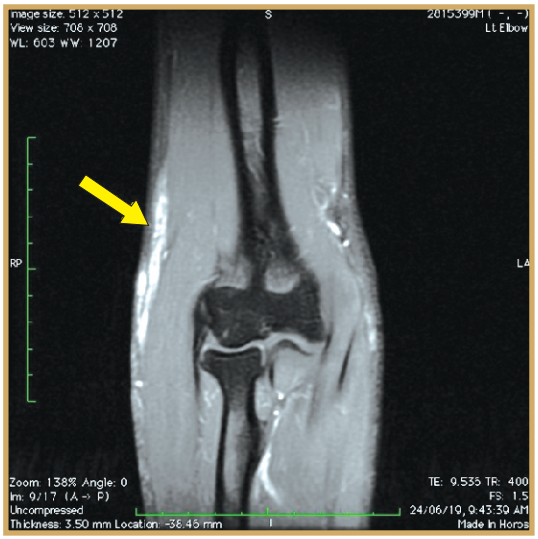

Fig. 3a : MRI (sagittal section) : Well defined ovoid branching focus of altered signal intensity (yellow arrow) continuous with nerve in the subcutaneous tissue on lateral aspect of distal third of arm.

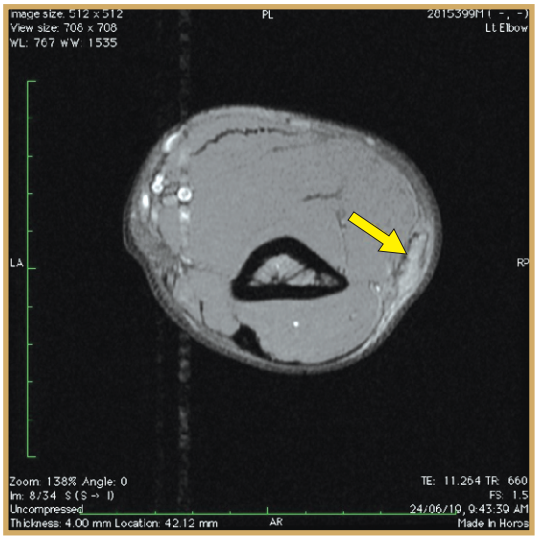

Fig 3b : MRI (axial section) : Well defined ovoid branching focus of altered signal intensity (yellow arrow) continuous with nerve in the subcutaneous tissue on lateral aspect of distal third of arm.

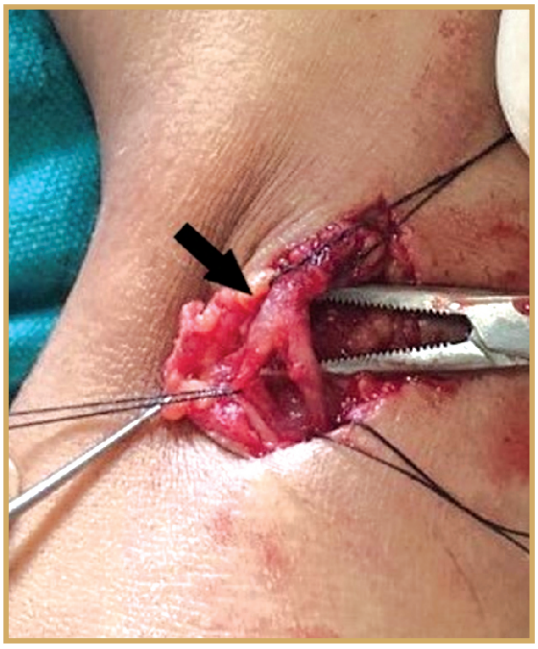

Fig 4a : Intraoperatively thickened lateral cutaneous nerve of forearm

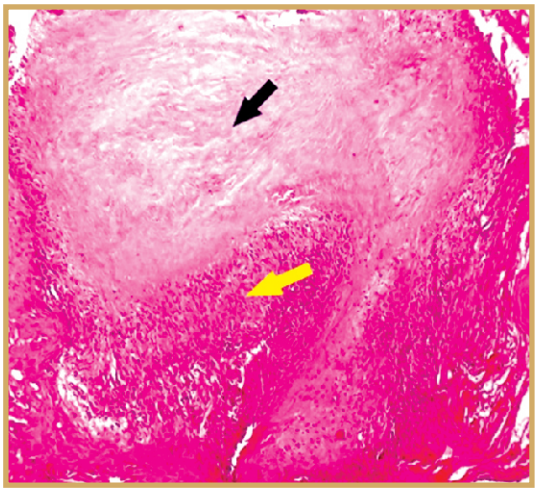

Fig 5 : Nerve biopsy: Hematoxylin & Eosin stain (40X) - large areas of necrotic tissue (black arrow) along with peripheral inflammatory cells comprising of lymphocytes and macrophages (yellow arrow)

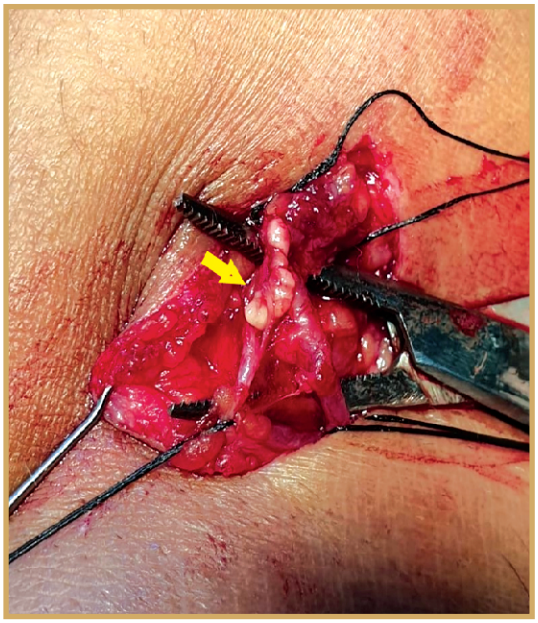

Fig 4b : Cheesy white caseous material expressed on nerve decompression

found to contain cheesy white matter (Figs. 4a and 4b). Nerve tissue, was sent for histopathological examination which revealed large areas of necrotic tissue along with peripheral inflammatory cells comprising of lymphocytes and macrophages. Many well-formed granulomas were seen (Fig. 5). ZN stain was negative. Patient responded well post nerve decompression and was continued on oral steroids for eight weeks in tapering doses. Presently patient is on 3 drugs MDT with significant reduction in size of leprosy patch and complete resolution of nerve abscess.

Discussion

Guide to Leprosy Control (WHO 1988) defines relapse as a patient who successfully completes an adequate course of MDT, but who subsequently develops new signs and symptoms of the disease either during the surveillance period (2 years for PB and 5 years for MB leprosy) or

thereafter (Sehgal et al 1996). Relapse in leprosy can be caused by insufficient treatment causing early relapse, and persistent mycobacteria leading to late relapse. Persisters are viable organisms fully susceptible to drugs but survive despite adequate treatment, probably because it is in a low or dormant metabolic state. Our patient fell into the criteria of early relapse possibly due to insufficient treatment thereby suggesting erstwhile 2 drug MDT regimen as per WHO recommendation in cases of PB leprosy for 6 months is not adequate in all patients.

Paucibacillary leprosy was traditionally treated with two drug MDT regimen containing Rifampicin and Dapsone for 6 months, since the introduction of WHO multidrug therapy (MDT) in 1982. Effectiveness of a drug regimen in any infectious disease is based on two important factors: incidence of relapse and amelioration of sign and symptoms. WHO has estimated a risk of relapse of 0.77% for MB and 1.07% for PB patients 9 years after stopping MDT (Kaimal & Thappa 2009). Grugni et al (1988) found relapse rate of 5.63% (17.5 per 1000 person years at risk) in PB leprosy). Various other studies have reported a higher relapse rates in PB patients using a shorter 2-drug regimen (Kumar et al 2012). So, it can be safely presumed that the occurrence of higher relapse rate in PB leprosy in comparison to MB leprosy is either due to misclassification or inadequate therapy. Hence, 3-drug regimen is a simple and a logical solution to reduce the incidence of relapse.

In concurrence with the same, the present WHO guidelines (2018) for treatment of leprosy recommends the same 3-drug regimen with rifampicin, dapsone and clofazimine for all leprosy patients, with a duration of treatment of 6 months for PB leprosy and of 12 months for MB leprosy (WHO 2018). The Guidelines development group (GDG) justify a 3-drug regimen for PB leprosy as there is evidence of better clinical outcomes with a 3-drug 6-month regimen compared with a 2-drug 6-month regimen (Rao et al 2009, Prasad et al 2005). An additional advantage of clofazimine addition is in reducing the persisting activity, incidence of reactions / relapses and neuritis, which is a serious complication leading to deformities (Katoch et al 1999, Nair 2018), thereby overall improvements in clinical outcomes (Katoch et al 1999) Since our patient had been treated with two drug MDT in the past and presented with clinical and histopathological features of relapse we restarted our patient on 3 drugs MDT.

Nerve abscesses are rare in leprosy with a prevalence of 1.3% in India (Gaur et al 1994). Abscesses have been reported in leprosy post completion of MDT (Agrawal et al 2005) commonly in ulnar nerve, but rarely reported in lateral cutaneous nerve of forearm. The lateral cutaneous nerve of forearm or lateral antebrachial cutaneous nerve is a branch of musculocutaneous nerve which passes behind the cephalic vein and divides into volar and dorsal branch. The volar branch supplies skin of lateral half of forearm and the dorsal half supplies the skin over lateral two thirds of dorso-lateral aspect of forearm. Our patient presented with nerve abscess in this cutaneous nerve after having been treated with WHO PB MDT, with clinical as well as histopathological evidence of relapse.

Pathogenesis of nerve abscess in leprosy is multipronged. Bacterial parasitization of the nerves in leprosy leads to the formation of granulomatous lesions due to anoxia produced by stretching, and pressure on the nerve secondary to inflammation and vascular damage. Caseous

necrosis in these nerve lesions occasionally coalesces to form a nerve abscess (cold abscess) particularly where the immunity is high as in PB spectrum of leprosy (Rai et al 2013). Furthermore, following multidrug therapy, an intense inflammatory response and subsequent damage to the peripheral nerves occurs, which may at times be intense enough to produce nerve abscesses (Salafia & Chauhan 1996).

Once diagnosed, a leprosy associated nerve abscess often requires surgical decompression and drainage of the abscess for possible restoration of nerve function to preventing the expansion of the inflammatory process to adjacent intact fibres (Abraham et al 1997). In our case nerve decompression was done as patient did not respond to oral steroids.

This case is being reported for two reasons: Firstly, lateral cutaneous nerve of forearm has been very rarely reported to develop nerve abscess and unless palpated for carefully, is likely to be missed. Secondly nerve abscess developing as a manifestation of relapse in a cutaneous nerve is rare. Treatment of leprosy should be continued till all signs of activity have subsided and clinical as well as histopathological regression has taken place. Regular follow-up (once in 6 months) of leprosy patients is necessary so that relapses and complications like nerve abscesses are detected early.

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