

# ISOLATED SUPERFICIAL PERONEAL NERVE LESION IN PURE NEURAL LEPROSY

## Case report

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**ABSTRACT** - Patients with leprosy may have only nerve involvement without skin changes. These cases are known as pure neural leprosy and can be seen in 10% of leprosy patients. Most patients have mononeuritic or multiple mononeuritic neuropathy patterns. The isolated lesion of the superficial peroneal nerve is uncommonly seen. We report a patient with involvement of this nerve in which there was no thickening of superficial nerves. The performed nerve biopsy showed inflammatory infiltration, loss of fibers and presence of *Mycobacterium leprae*. We believe that in prevalent leprosy countries we should take in mind the possibility of isolated pure neural leprosy in some patients without skin lesion. In these cases the diagnosis of leprosy is impossible on clinical grounds and nerve biopsy is mandatory.

**KEY WORDS:** leprosy, hanseniasis, superficial peroneal nerve, nerve biopsy, pure neural leprosy.

### **Acometimento isolado do nervo peroneiro superficial na lepra neurítica pura: relato de caso**

**RESUMO** - Em alguns casos de lepra podemos encontrar acometimento de nervos periféricos sem manifestações cutâneas. É a denominada lepra neurítica pura que pode ocorrer em até 10% dos casos desta doença. Na maioria das vezes predominam quadros de mononeuropatia ou mononeuropatia múltipla. O acometimento isolado do nervo peroneiro superficial tem sido pouco relatado. Referimos a uma paciente com acometimento isolado deste nervo sem hipertrofia de troncos nervosos. A biópsia realizada mostrou presença de infiltrado inflamatório, perda moderada de fibras e presença do *Mycobacterium leprae*. Acreditamos que, em países onde a lepra é endêmica, diante de acometimento isolado de nervos periféricos, deve-se pensar na possibilidade de se tratar da forma neurítica pura desta moléstia, mesmo em pacientes sem alterações dermatológicas. Nestes casos o diagnóstico só será possível com a realização de biópsia de nervo superficial.

**PALAVRAS-CHAVE:** lepra, hanseníase, nervo peroneiro superficial, biópsia de nervo, lepra neurítica pura.

Leprosy is one of the most common peripheral nerve diseases in the world. It usually affects the skin and the nerves. It is caused by the *Mycobacterium leprae*, an acid-fast bacillus. Although the prevalence of the disease is decreasing, leprosy continues to be a major cause of infective neuropathy in tropical and subtropical countries. Patients with leprosy may have only nerve involvement without obvious primary skin lesions. The so-called pure neuritic form is a well-recognized clinical entity, comprising 4 to 10% of patients with leprosy<sup>1,2</sup>. In this pure neuritic form, the ulnar and the common peroneal nerves are the most frequently affected nerves<sup>1,3</sup>. Rare cases with superficial peroneal nerve involvement are referred in leprosy pure neuritic form<sup>1</sup>.

We report a patient with superficial peroneal nerve com-

promise without nerve hypertrophy, and also without skin changes.

### **CASE**

A 62 year-old woman was referred with progressive paresthesia in the outer aspect of the left leg. She had been well until three years early when she began to feel progressive paresthesia and numbness over the outer side of the left leg. She took enalapril during 5 years for hypertension. There is no history of alcoholism. Diabetes mellitus type 2 was diagnosed four months before examined in our Service. Her family history disclosed no cases of leprosy or other cutaneous disorders.

On physical examination patient appeared well. Dermatological examination was normal. Neurological examination showed sensory loss (touch, pain and temperature) of the lateral aspect of the left

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Table 1. Sensory nerve conduction study.

	Distal latency (ms)	Amplitude ( $\mu$ v)	Nerve conduction (m/s)
<i>Upper limb</i>			
R median	3	22	53
L median	3.1	26	51
R ulnar	2.8	23	51
L ulnar	2.7	19	50
<i>Lower limb</i>			
R sural	3.4	18	43
L sural	3.5	13	44
L Superficial peroneal	0	0	
R Superficial peroneal	0	0	

R, right; L, left.

lower half of the leg and dorsum of the left foot. The proprioceptive sensation was normal. Tendon reflexes were present. No other abnormality was observed in neurological examination. No superficial nerve with hypertrophy was palpable. The oral glucose tolerance test showed: fasting glucose 103 mg%, 2 hours following 75 g of oral glucose: 143 mg%, glycosylated haemoglobin: 8,9 % (normal: 5.5 - 8.5 %). Other blood tests and the hematological values were within normal ranges. The lepromine test was not performed. Motor nerve conduction study was normal. The sensory action potential of both superficial peroneal nerves was not elicited (Table 1).

We performed a left superficial peroneal nerve biopsy in the lower one third of the leg. The specimen was fixed in a 3% glutaraldehyde, stained with hematoxylin & eosin (H&E), and Gomori's trichrome. Congo red staining for amyloid and Wade's staining for bacilli identification were performed. Semi-thin sections were stained with toluidine blue. Optic microscopy showed: 1) intense inflammatory infiltrates consisting of macrophages, plasma cells and lymphocytes mainly in the endoneurial space (Fig 1a and b); 2) presence of vacuolated "foamy cells"; 3) mild fibrosis of endoneurium, perineurium and epineurium; 4) asymmetrical partial loss of nerve fibres (Fig 2a); 5) large number of bacilli in the endoneurium, particularly in endothelial cells and Schwann cells (Fig 2b).

## DISCUSSION

The common peroneal nerve divides at the neck of the fibula in two branches, the superficial (Fig 3) and the deep peroneal nerves. The superficial peroneal nerve descends along the shaft of the fibula, in the lateral compartment of the leg, close to the peroneal muscles that it supplies<sup>4</sup>. When it is affected there is a sensory loss covering the lateral calf, the lateral malleolus, the dorsum of the foot and the medial three or four toes. The superficial peroneal nerve is involved mainly by entrapment or direct injury. In most instances the lesion is located distal to the point of innervation of the peroneal mus-

cle and the symptoms are purely sensory<sup>4</sup>. It is doubtful if in diabetes mellitus peripheral neuropathy can appear as a mononeuropathy. Isolated lesion of the superficial peroneal nerve has never been directly related to this metabolic disease.

The high incidence of pure neural leprosy justifies its inclusion under a separate subgroup<sup>3</sup>. Mononeuritis is the most common presentation, and the ulnar being the most involved nerve<sup>1-3,5-7</sup>. Other nerves may be involved but very rarely. The other involved nerves are the common peroneal nerve, the median nerve, the radial and the facial nerves<sup>1,3</sup>. Taking into account twelve patients with pure neuritic leprosy, Uplekar and Antia<sup>8</sup> concluded that the pure mononeuritic form is much more common than the pure polyneuritic form. Stalwar et al.<sup>9</sup> studied 42 cases of neuritic leprosy. All of them had mononeuritic or multiple mononeuritic involvement. The ulnar was the most common involved nerve followed by the lateral popliteal nerve. No case with superficial peroneal nerve involvement was identified in this series. Mahajan et al.<sup>3</sup> described 179 patients with pure neuritic leprosy. In 31 of these patients there was an involvement of only one nerve. The most common neuropathic presentation was the involvement of two nerves in the same extremity. These authors did not identify an isolated superficial peroneal involvement although the ulnar and the common peroneal were together the most affected nerves. Girdhar<sup>1</sup> referred to 214 cases of pure neuritic leprosy studied till 1996 by several authors. The ulnar and the common peroneal were the two most frequently affected nerves. The superficial peroneal was one of the less involved nerves. Although not a commonly involved nerve Vaidyanathan and Vaidyanathan<sup>10</sup> performed biopsy of this nerve in 54 patients with superficial peroneal nerve thickening. Five of these patients had symptoms and signs related to skin or nerve

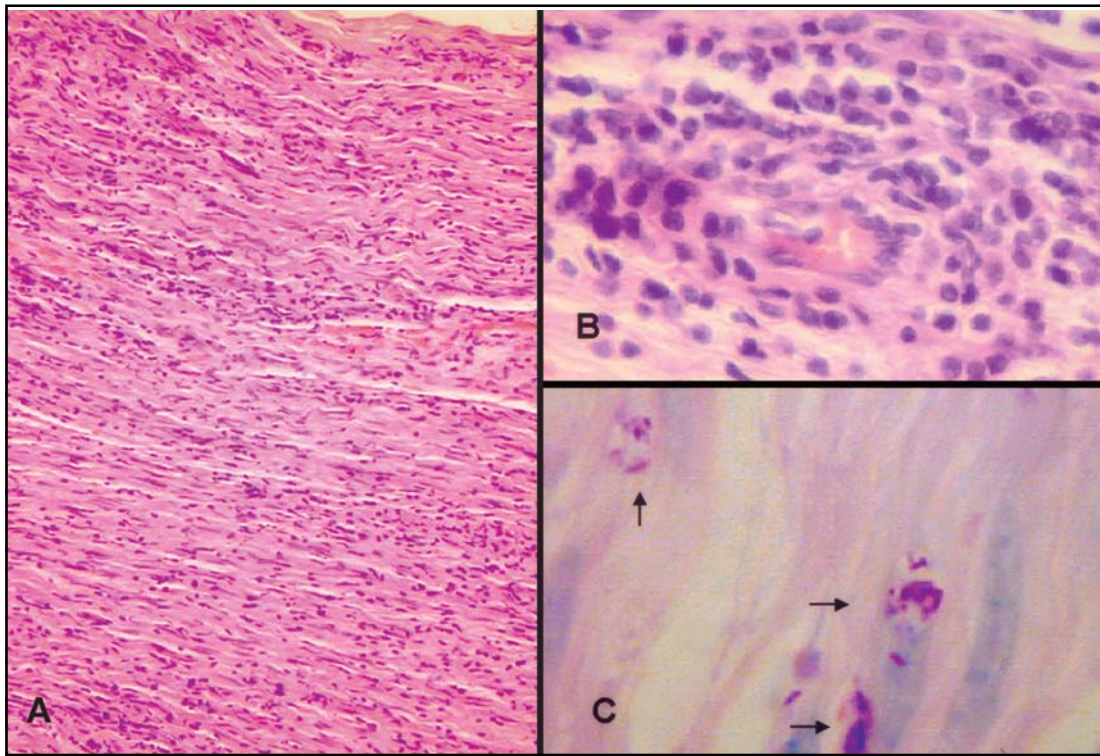


Fig 1. Superficial peroneal nerve biopsy. A) Intense inflammatory reaction involving endoneurium, H&E X 10. B) Lymphocytic vasculitis affecting epineurial blood vessels, H&E X 40. C) Presence of bacilli (arrows), Wade X 100.

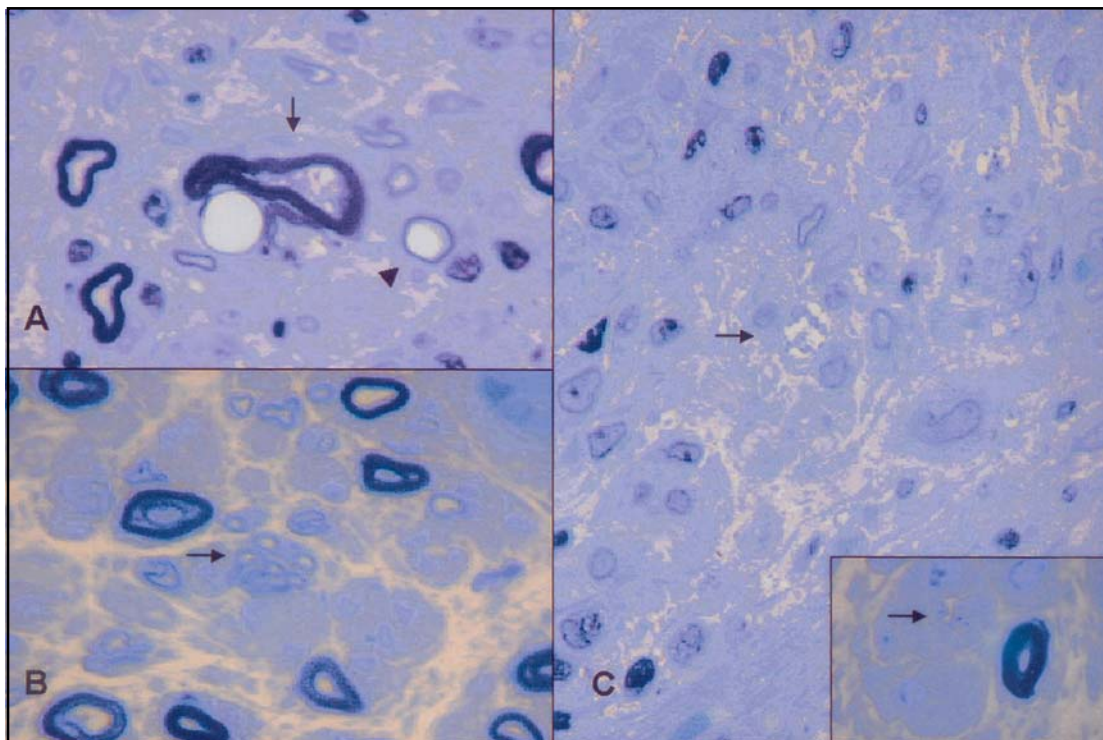


Fig 2. Superficial peroneal nerve biopsy. A) Transverse section showing partial loss of myelinated fibers with axonal degeneration (arrow) and remyelination (arrowhead). B) Presence of regenerative cluster (arrow). C) Total loss of myelinated fibers. Bacilli were present in "foamy cells" (arrows). Toluidine blue X 40. Insert: a normal myelinated fiber near many bacilli (X 100).

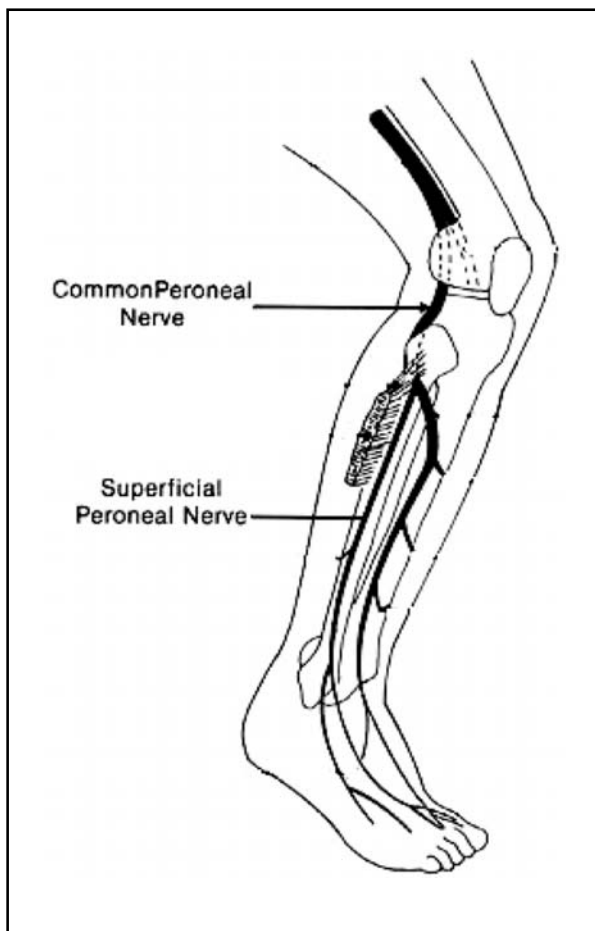


Fig 3. Superficial peroneal nerve.

damage, but 49 were symptom free. There was irrefutable histopathological evidence of leprosy in 50% of the biopsies.

In our patient there was no thickened nerve. The number of enlarged nerves in neuritic leprosy varies among different series. Pannikar et al.<sup>5</sup> reported 17 cases of pure neuritic leprosy, 13 of them (76.5%) having clinically enlarged nerves. In the 214 previously reported cases of neuritic leprosy revised by Girdhar<sup>1</sup> nerve enlargement findings varied from 33 to 83%. Recently Benedetti et al.<sup>11</sup> performed Magnetic Resonance Image (MRI) in one case of leprosy and showed prominent diffuse thickening in various nerves. Although these findings are nonspecific, MRI could help in the diagnosis of hypertrophic neuropathies.

Motor nerve conduction study was normal in all examined nerves in the upper and lower limbs of our patient. The sensory nerve conduction study was almost normal, exception was an unexcitable superficial peroneal nerve in both legs. This electrophysiological finding is suggestive of an isolated peroneal nerve damage but is nonspecific.

In most cases of primarily neural leprosy the diagnosis can-

not be firmly established on clinically grounds alone. Only the nerve biopsy could establish the exact diagnosis. We have studied the superficial ulnar nerve biopsy in the dorsum of the hand in 20 cases of isolated ulnar palsy in leprosy<sup>7</sup>. We found unequivocal pathological evidence of leprosy in all of them. In 77 patients with the diagnosis of primary neuritic leprosy Jacob and Mathai<sup>12</sup> performed cutaneous nerve biopsy near to the site of neurological deficit. Half of them had leprosy confirmed. Kaur et al.<sup>6</sup> stated that 35 out of 37 enlarged nerves of patients with suspected leprosy presented lepromatous infiltrates in nerve biopsy. In 67 suspected patients Jardim et al.<sup>13</sup> confirmed the diagnosis of leprosy in 73% by a nerve biopsy. In some cases without neurological symptoms and signs the nerve biopsy may reveal infiltration of the nerve by the *Mycobacterium leprae*<sup>14</sup>. Superficial sensory nerve biopsy is a relative simple office procedure and is an important tool for the diagnosis of primary neuritic leprosy.

Histological classification of a nerve biopsy specimen in leprosy is based chiefly on the character of the cellular infiltrate and the bacillary load. As most cases of pure neural leprosy consist of mononeuropathy or multiple mononeuropathy the nerve histopathology pattern is toward the tuberculoid leprosy pole<sup>9</sup>. In the polyneuritic form the histological findings are consistent with the lepromatous features containing many bacilli<sup>15,16</sup>. In addition, as in our case, there is evidence that even the mononeuritic form may reveal changes consistent with lepromatous leprosy<sup>1,5</sup>.

We conclude that in cases of mononeuropathy without a known etiology and without skin changes suggestive of leprosy it is mandatory to perform a superficial nerve biopsy to confirm the diagnosis of pure neural leprosy, mainly in developing countries where leprosy is prevalent.

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