

Surgical treatment of type I neuritis in a teenage boy with borderline tuberculoid leprosy

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ABSTRACT

Exacerbation of the immune response against *Mycobacterium leprae* can lead to neuritis, which is commonly treated via immunosuppression with corticosteroids. Early neurolysis may be performed concurrently, especially in young patients with a risk of functional sequelae. We report the case of a young patient experienced intense pain in the left elbow one year after the treatment of tuberculoid-tuberculoid leprosy. The pain was associated with paresthesias in the ulnar edge and left ulnar claw. After evaluation, the diagnosis was changed to borderline tuberculoid leprosy accompanied with neuritis of the left ulnar nerve. Early neurolysis resulted in rapid reduction of the pain and recovery of motor function.

Keywords: Borderline leprosy. Neuritis. Leprosy therapy. Minor surgical procedures. Leprosy surgery.

INTRODUCTION

Leprosy has become a curable disease after the introduction of multidrug therapy (MDT). Nevertheless, deformities caused by *Mycobacterium leprae* (*M. leprae*) infection are still common in older adults and elderly patients in some developing countries. These complications, however, are rare in young patients, and when present, they suggest incorrect or incomplete diagnosis and inadequate or poor medical care⁽¹⁾⁽²⁾.

Deformities related to neural damage are directly associated with cellular immunity, presence of Type I reactions (TIRs), and MDT⁽³⁾⁽⁴⁾. Exacerbation of the immune response against *M. leprae* may lead to neuritis (also called TIR in the nerves, or reverse reaction) characterized by substantial intraneural edema, granulomatous reactions, pain, and thickening of the nerve path⁽³⁾.

The conventional treatment for neuritis includes immunosuppression with corticosteroids⁽⁴⁾. Neurolysis has been described as the most effective therapeutic option in cases with no response after four weeks of therapy, for subintractant chronic neuritis, or if an abscess is present⁽⁴⁾⁽⁵⁾. Early neurolysis is an alternative therapy that should not be neglected and may be performed concurrently with corticosteroids administration, especially in young patients with a risk of functional sequelae or significant nerve involvement.

CASE REPORT

A 14-year-old boy was previously diagnosed with tuberculoid-tuberculoid leprosy at another facility and received treatment with paucibacillary MDT. He had had a two-year history of two anesthetic hypopigmented patches. Intense pain in the left elbow associated with paresthesias in the ulnar edge and left ulnar claw developed one year after the completion of drug treatment. An abscess and left ulnar nerve neuritis were suspected, and he was referred to our leprosy reference service.

On admission the patient had left ulnar claw. Palpation of the left ulnar nerve revealed thickening associated with significant pain. Four xerodermichypochromic, anesthetic patches with imprecise borders were distributed on the elbow, thigh, back of the left hand and right palm (**Figure 1A, B, C, D, E and F**).

The results of routine complementary tests were as follows: a) Mitsuda reaction 14mm; b) phenolic glycolipid-1 (PGL-1) antibodies 0.029 – nonreactive; c) sensory motor test decreased sensitivity in the area of the left ulnar nerve (monofilament test: black color); and d) electromyography pronounced predominantly axonal mononeuropathy of the left ulnar nerve across the elbow with signs of inflammatory neuropathy and reduction of the left ulnar nerve conduction (17.3m/s).

Biopsy of the skin lesion revealed borderline tuberculoid leprosy in remission, negative acid-fast bacilli smear, and associated TIR (**Figure 2A and B**). Ultrasonography of the left elbow ruled out abscess and showed profound neural thickening (11.9 x 7.3mm) with diffuse increased vascularity, suggestive of neuritis. Based on these results, the patient was diagnosed

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with borderlinetuberculoid leprosy accompanied with neuritis of the left ulnar nerve. We proceeded with multibacillary MDT, administration of corticosteroids in immunosuppressive doses (prednisone 1mg/kg/day) and early neurolysis.

Surgery revealed a stiffened left ulnar nerve with increased diameter and a thickened perineurium. We proceeded with an incision at the Osborne's ligament and on the fascia of the triceps tendon and flexor carpi ulnaris. Longitudinal incision of the epineurium revealed enlarged fibrous bundles without necrosis (**Figure 3A, B, C, and D**). The corticosteroid therapy was continued postoperatively for six months^{(4) (5)}.

The patient experienced significant improvement in pain during the early postoperative period, reduction in the ulnar claw

(**Figure 1F**) two months after the surgery, improvement in motor conduction (43.3m/s), and a slight improvement in sensitivity.

DISCUSSION

The current leprosy classification is based on the number of skin lesions and affected nerves. In particular, cases with up to five skin lesions and/or one damaged peripheral nerve are classified as paucibacillary leprosy, whereas those with more than five skin lesions and/or more than one peripheral nerve involvement are classified as multibacillary leprosy. Although this clinical classification has some merit, its usage may result

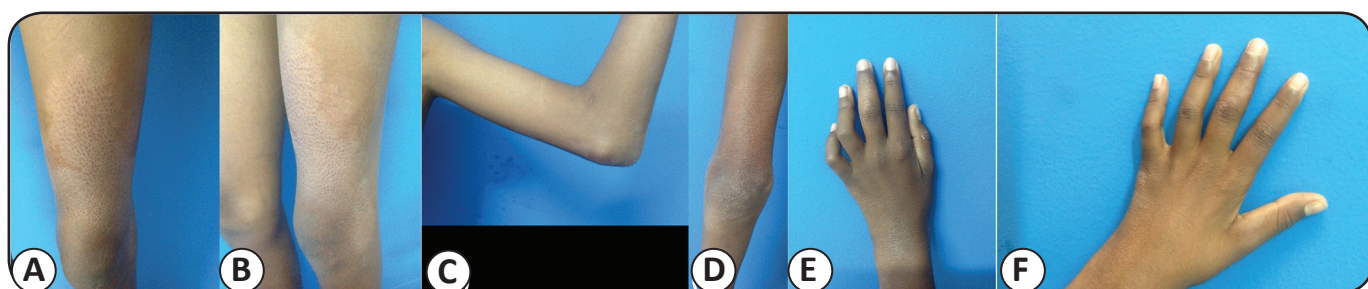


FIGURE 1 - A and B: A hypochromic and xerodermic patch with imprecise borders and definite sensory loss on the left lower limb. C and D: A hypochromic and xerodermic patch with a loss of sensitivity on the left elbow and a tumor on the medial epicondyle. E: The left hand with flexible ulnar claw. F: The reduction of ulnar claw two months after neurolysis and immunosuppressive therapy with corticosteroids.

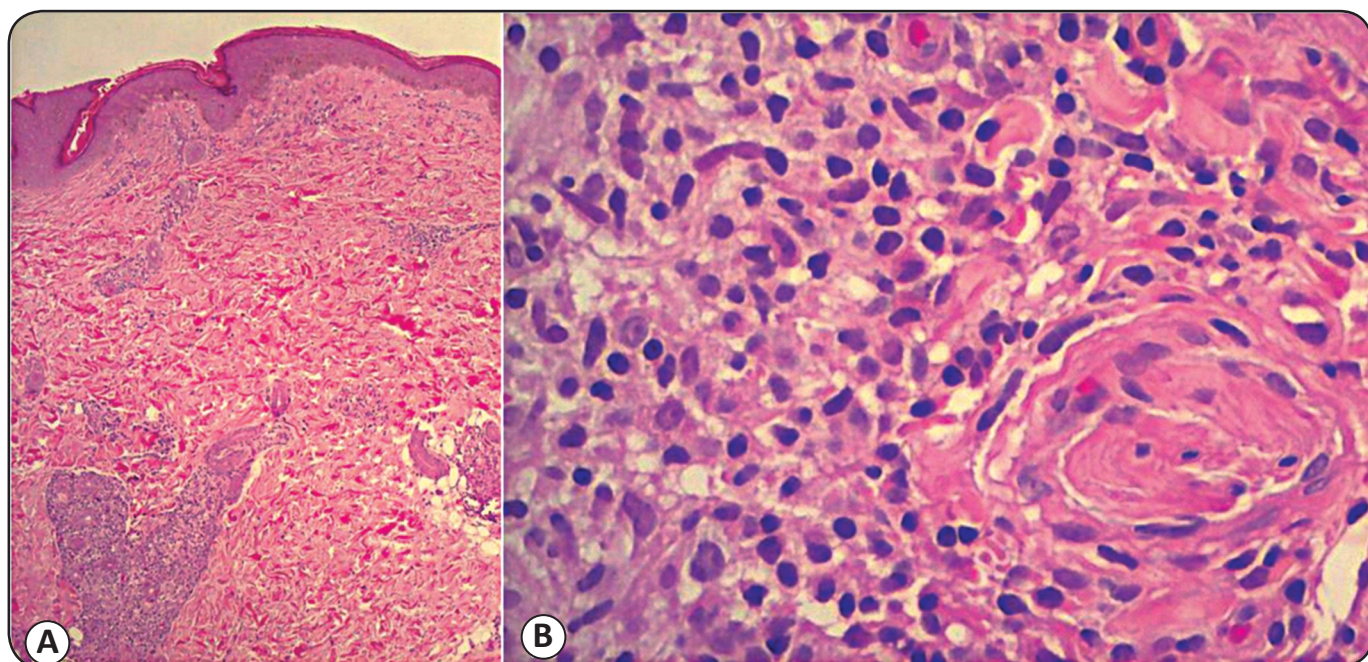


FIGURE 2 - A: A microphotograph of skin showing a multifocal, superficial deep tuberculoid granulomatous reaction (hematoxylin and eosin, original magnification x40). B: A higher magnification photograph showing the granulomatous epithelioid reaction (left side) with perineural edema surrounded by monocytes and lymphocytes and thickening of the perineurium (right side) typical of reactional borderline tuberculoid leprosy (hematoxylin and eosin, original magnification x200). The biopsy bacilloscopy index was negative on Faraco-Fite staining.

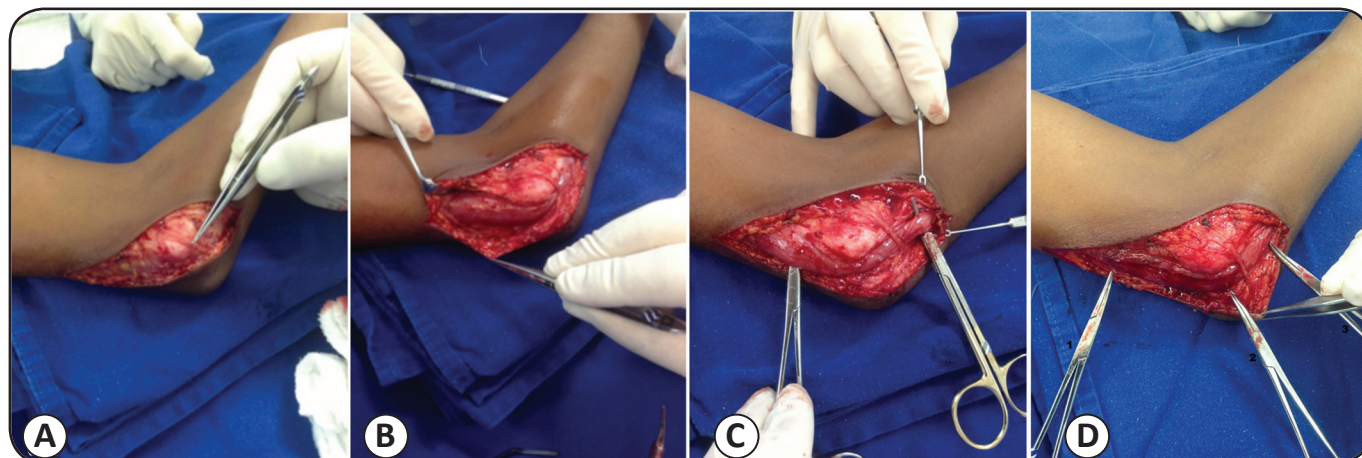


FIGURE 3 - A: A perioperative image of the medial region of the elbow with exposure of the thickened sensory nerve branch. **B:** Exposure of the ulnar nerve (about 5 times thicker than usual) above and below the elbow. The ligament of Osborne and the ulnar carpal flexor muscle were excised. **C:** The ulnar nerve after the dissection of the *flexor carpi ulnaris*. **D:** Exposure of the thickened ulnar nerve. The tips of the tweezers indicate the points of extrinsic nerve compression: 1) the fascia of the triceps; 2) the ligament of Osborne; and 3) the ulnar head of the *flexor carpi ulnaris*.

in establishment of ineffective treatments, primarily because of errors in disease categorization⁽¹⁾⁽²⁾.

The presence of bacillary antigens initiates TIR in the nerves, causing nerve damage⁽³⁾⁽⁶⁾⁽⁷⁾. The inflammatory process leads to intraneural edema, resulting in pain and thickening⁽³⁾⁽⁵⁾⁽⁸⁾, which causes compression in the *vasa nervorum* and subsequent ischemia of the nerve⁽¹⁾⁽³⁾⁽⁶⁾. Another consequence of the inflammatory process is intrinsic compression exerted by the epineurium and extrinsic compression exerted by the adjacent nerve structures. In the ulnar nerve, such critical structures are the Osborne's ligament, fascia, muscles of the triceps, and the tendon of the *flexor carpi ulnaris*⁽⁵⁾⁽⁶⁾⁽⁹⁾.

The current recommendations for the management of neuritis in leprosy patients suggest immunosuppressive therapy with corticosteroids for at least four weeks to inhibit the cellular immune response and the consequent destruction of nerve cells⁽⁶⁾⁽⁷⁾. Neurolysis is usually indicated in cases with neural abscess, intermittent neuritis, and poor or absent response after four weeks of corticosteroids use⁽⁵⁾⁽¹⁰⁾⁽¹¹⁾.

In the present report, early surgical intervention was performed because of the significant compression of the ulnar nerve associated with delayed motor conduction, reduced sensitivity and the presence of ulnar claw. In patients with substantial nerve enlargement accompanied with pain and rapid loss of function, the use of isolated clinical treatment can be controversial. A randomized trial comparing medical and medico-surgical approaches in leprosy patients reported significantly better results for the latter in terms of reducing pain and limiting nerve involvement⁽¹²⁾. Thus, minimizing the time required to reduce inflammation and intraneural edema may be critical for the prevention of subsequent deformities. Other authors agree that in some cases steroid therapy alone is ineffective in relieving pain and reducing nervous involvement and damage⁽¹¹⁾⁽¹²⁾.

In this report, there was a significant positive response to the surgical treatment combined with corticosteroids, including the rapid reduction of pain and recovery of motor function. The limited improvement in sensitivity in the region of the

ulnar nerve was probably a consequence of previous damage to the sensory nerve fibers. It is worth mentioning that surgical intervention was considered only after a thorough clinical examination. Importantly, treatment of such cases should be performed in a specialized leprosy reference service.

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