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Case-based Review

An unusual case of tenosynovitis in Hansen's disease

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Abstract

Tenosynovitis is an infrequent presentation of Hansen's disease. It may occur during the natural course of disease or treatment as part of type 1 reaction or rarely may be the presenting complaint. We report a case of tenosynovitis of bilateral wrist joints who after being ineffectively treated by an orthopedician as well as rheumatologist for several months and was finally diagnosed as a case of Hansen's disease (borderline lepromatous) in type 1 reaction with excellent response to multidrug therapy and tapering doses of systemic steroids.

Keywords: Hansen's disease, leprosy, rheumatological manifestations, tenosynovitis

Introduction

Leprosy may present with myriad of manifestations mimicking dermatological, medical, neurological, and rheumatological diseases.[1] This often causes diagnostic dilemma, leading to delay in institution of treatment. Rheumatological involvement as a presenting feature ranges from 1% to 70%.[2] Tenosynovitis is an unusual presenting feature of Hansen's disease.[3]

Case Presentation

A 35-year-old male presented to an orthopedician with painless swelling and pain over the dorsa of both the wrist joints of 3 weeks duration, who managed him conservatively with analgesics without much relief. He then consulted a rheumatologist clinic where some radiological and blood tests were carried out, and he was started on monthly injections of 40 mg mL^{-1} methylprednisolone acetate into both the wrist joints and oral analgesics. He showed gradual improvement to the therapy over the next 4 months. However, over the course of next 2 weeks, he developed asymptomatic some red plaques over his face, neck, trunk, elbows, forearms, hands, and feet; for which, he was referred to dermatology outpatient department. Clinical examination revealed two well-defined, nontender, soft, fluctuant, cystic swellings roughly measuring 5 × 4 cm over the dorsa of each of the wrist joints, which moved with flexion and extension (Figure 1). Dermatological examination revealed multiple well-defined discrete erythematous plaques involving his face, trunk, and extremities (Figure 2). The lesions were hypoaesthetic to temperature, fine touch, and pain. Bilateral ulnar nerves and common peroneal nerves were uniformly thickened and nontender. Ultrasound imaging of the swelling of wrist joints revealed mild synovial thickening and effusion, involving the tendon sheaths of extensor digitorum longus bilaterally and extensor carpi radialis brevis of right wrist and extensor carpi radialis longus and brevis of left wrist (Figure 3). Modified Ziehl Neelsen (ZN) stain on slit skin smear for lepra bacilli was negative. The aspirate from swelling of both the wrists showed inflammatory cells comprising mainly of lymphomononuclear cells with few neutrophils and negative modified ZN stain for lepra bacilli. Skin biopsy from an erythematous plaque over the trunk revealed few ill-formed epitheloid granulomas in the dermis along with mild perivascular lymphonuclear and histiocytic inflammatory infiltrate consistent with type 1 reaction in a case of Hansen's disease (Figure 4). He was diagnosed as a case of Hansen's disease (borderline lepromatous) in type 1 lepra reaction in the form of erythema of the skin lesions with bilateral tenosynovitis of extensor tendons of the wrist joints. The patient was started on multidrug therapy (MDT) comprising of dapsone 100 mg once daily, cap clofazimine 50 mg daily, and 300 mg monthly and cap rifampicin 600 mg monthly with tapering doses of oral prednisolone starting at 1 mg kg^{-1} body weight. The swelling over both the wrist joints disappeared completely in 2 months (Figure 5), and the skin lesions started healing with post-inflammatory hyperpigmentation (Figure 6). The patient is presently under follow-up, and no recurrence of joint or skin lesions has occurred after stopping oral prednisolone and 6 months of MDT.

Discussion

Though rheumatological presentations in leprosy especially in the setting of reactions are not unknown, tenosynovitis is an extremely unusual presentation.[4] A meticulous clinical examination for

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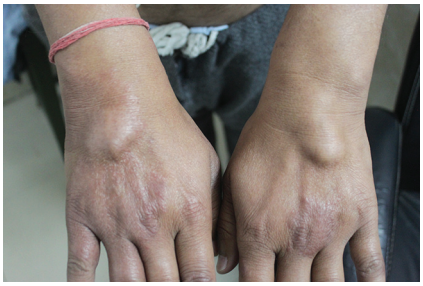

Figure 1. Two well-defined, nontender, soft, fluctuant, cystic swellings roughly measuring 5 × 4 cm over the dorsa of each of the wrist joints, which moved with flexion and extension. Closer examination also reveals 2× erythematous to hyperpigmented, hypoaesthetic plaques on the dorsum of both hands; the plaque on the right hand is greater in size than the left.

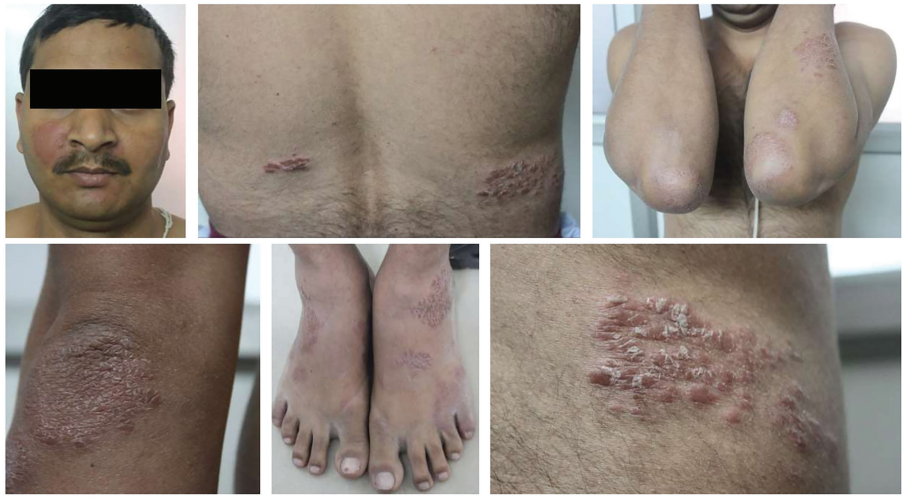

Figure 2. Multiple well-defined discrete erythematous hypoaesthetic plaques on face, trunk, and extremities.

skin and neural involvement looking for other clues for Hansen's disease may establish the underlying diagnosis in a patient seeking referral for rheumatologic symptoms. Tenosynovitis in Hansen's disease generally occurs with type 1 lepra reaction and presents as swellings over dorsa of hands and/or feet.[5] Other causes of tenosynovitis are rheumatoid arthritis, psoriatic arthropathy, gout, repetitive strain injury, infective organisms like *Staphylococcus aureus*, Methicillin-Resistant Staphylococcus aureus (MRSA), *Staphylococcus epidermidis*, beta-hemolytic *Streptococcus*, *Pseudomonas aeruginosa*, etc.[6] Autoimmune complexes, deposition of crystalline substances, overuse, and other above etiologies affect the tendon or the synovium, resulting in inflammation and subsequent fibrosis and loss of function.[7] Tenosynovitis in leprosy is caused by the hematogenous spread of the bacilli followed by trapping of the bacillary antigens in synovial lining of the tendon sheath giving rise to inflammatory cascade.[8] Infectious tenosynovitis progresses over a series of stages—first

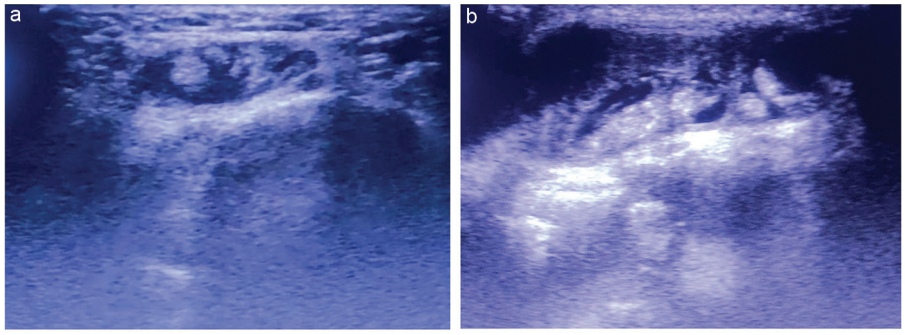

Figure 3. a,b. Ultrasound imaging of the swelling of (a) right and (b) left wrist joints: mild synovial thickening and effusion involving the tendon sheaths of extensor digitorum longus bilaterally and extensor carpi radialis brevis at right wrist and extensor carpi radialis longus and brevis at left wrist.

Main Points

- Tenosynovitis is a rare presentation of Hansen's disease.
- Tenosynovitis in Hansen's disease occurs as part of Type-1 reaction.
- It appears as fluctuant cystic swellings on dorsa of wrists and ankles.
- The management of choice is systemic or intralesional steroids.
- The management of choice is systemic steroids in addition to multidrug therapy for Hansen's disease.

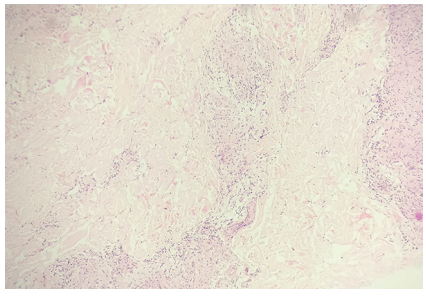

Figure 4. Skin biopsy from an erythematous plaque over the trunk revealed few ill-formed epitheloid granulomas in the dermis along with mild perivascular lymphonuclear and histiocytic inflammatory infiltrate consistent with type 1 reaction in a case of Hansen's disease (H and E, ×20).

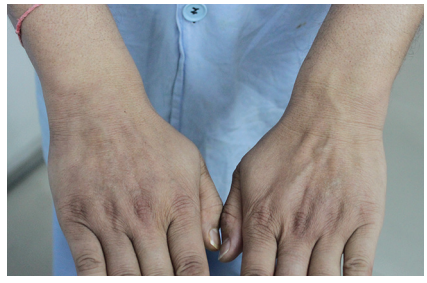

Figure 5. Resolution of tenosynovitis after 2 months of multidrug therapy and tapering doses of systemic steroids.

stage presents with distension of the tendon sheath complex, the second stage involves conversion of the distended fluid to purulent, and the third stage comprises of necrosis and destruction.[9] Our case later in the course of disease developed cutaneous type 1 reaction in the form of erythema of the skin lesions of leprosy. Sudden appearance of erythematous lesions over body may be explained by previously unknown subclinical lesions, which could have been manifested for the first time in type 1 reaction as reported in literature earlier.[10–12] Systemic corticosteroids along with MDT remain the cornerstone in the management of tenosynovitis of leprosy.[13] Our case highlights a rare

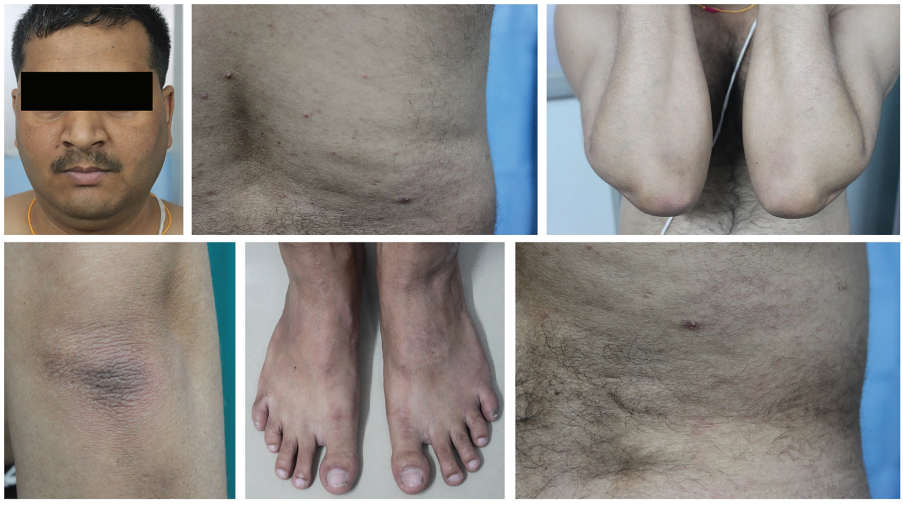

Figure 6. Resolution of tenosynovitis after 2 months of MDT and tapering doses of systemic steroids.

initial presentation of leprosy as tenosynovitis alone, mimicking a rheumatological disorder causing a diagnostic dilemma. The appearance of red raised lesions due to type 1 cutaneous reaction later in the course of disease helped suspect the diagnosis of Hansen's disease and starting of MDT with steroids leading to resolution of symptoms. The case is presented for its unusual and infrequent nature.

Informed Consent: Written informed consent was obtained from the patient who participated in this study.

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