CASE REPORT

ENL developing 10 years after leprosy treatment masquerading as atypical-mycobacterial infection

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Learning point for clinicians

Type II lepra reactions can occur after a long interval post therapy, which may span a decade as in this case. Skin lesions and lymphadenopathy can be the presenting features and would need careful exclusion of atypical mycobacterial infections. Modified Ziehl-Neelson stain is helpful in staining both these bacilli.

Case report

A 35-years-old gentleman presented with continuous, moderate grade fever for 7 days preceded by multiple erythematous, papulonodular lesions on the thighs, face and trunk, along with enlarged, discrete, and tender, cervical and inguinal lymph nodes. (Figure 1a) The symptoms persisted despite intake of antihistaminics. There were no thickened nerves or hypoesthetic skin lesions.

Aspirate from the cervical nodes revealed numerous foamy histiocytes and neutrophils on a necrotic background. No granulomas or atypical cells were identified. Ziehl-Neelson (ZN) stain showed hazy-red equivocal staining in the histiocytes. (Figure 1b) A possibility of atypical mycobacterial infection was raised. Routine blood investigations including hemogram and tests for Hepatitis B and C were normal. Erythrocyte sedimentation rate (ESR) was elevated (104 mm in first hour). Testing for human immunodeficiency virus by enzyme-linked immunosorbent assay was non-reactive on duplicate tests.

Additional smears from the cervical node aspirate were stained with a modified ZN stain (using 5% sulfuric acid decolorization). This revealed numerous rod-shaped bacilli reminiscent of lepra bacilli in globi. The bacilli also stained positive on Gomori stain. Based on the aspirate smear findings and the clinical scenario of fever, generalized lymphadenopathy, and raised ESR, a possibility of lepra reaction was considered. The patient was interviewed again and on direct questioning revealed a remote past history of leprosy about 10 years back when he had developed multiple hypoesthetic skin lesions. He had taken medication for the same, for 6 months, and defaulted further treatment. He had been asymptomatic for the past 10 years till the current episode, which in light of the previous history and cytology was a Type-II lepra reaction. No history of local trauma or contact with contaminated water was present.

The patient was started on multi-drug treatment (MDT) as per WHO guidelines, supplemented with steroids, and closely monitored for any worsening. The fever, skin lesions and enlarged lymph nodes subsided within a fortnight.

Discussion

The current case highlights several little known aspects of leprosy in reaction. Type II reactions or erythema nodosum leprosum (ENL) affect patients with lepromatous leprosy. The deposition of circulating immune complexes in the tissues results in inflammatory lesions in the skin, nerves, joints, testes, sclera and rarely lymph nodes.¹ A large cohort study including 2053 leprosy patients from India showed an incidence of ENL of 0.2%.² Type II reactions usually cluster during the first year of initiation of MDT or rarely occur in untreated patients as the initial presentation of leprosy.³ The occurrence...
of ENL after 10 years of treatment in our case is unique. In another study from India, late onset ENL occurring 2 years or more after release from treatment was seen to occur in 3% of multibacillary patients with rare cases presenting upto 8 years later. Atypical mycobacterial infection and lepra reaction may present with cervical lymphadenopathy and papulonodular skin lesions; Mycobacterium marinum and Mycobacterium chelonae, both, are known to cause painless papular skin lesions. Aspirates from lesions or nodes infected by atypical mycobacteria and lepra reaction may show similar morphology and ZN staining using 25% acid decolorization, as used for Mycobacterium tuberculosis, may not stain all atypical mycobacteria. (3) Use of 5% acid decolorization to stain lepra bacilli and atypical mycobacteria thus becomes essential in such cases.

We present a rare case of ENL developing 10 years after intake of MDT. The presentation as generalized lymphadenopathy and skin lesions entails the careful exclusion of atypical mycobacterial infection. Utilization of 5% acid decolorization and a positive past history of partially treated leprosy, reinforced by prompt response to MDT, clinched the diagnosis in our case. Such late onset ENL should be kept in mind by physicians for apt timely diagnosis and appropriate treatment.

Conflict of interest: None declared.

References