A Rare Presentation of Histoid Leprosy: A Case Report

PS Mohapatra¹, R Ray², M Khandare³

Histoid Leprosy is an expression of multibacillary leprosy, characterized by typical cutaneous and subcutaneous nodules and plaques present over an apparently normal skin and histopathologically the predominance of spindle-shaped cells or polygonal cells and unusually large number of acid fast bacilli. We report a case of histoid leprosy which was insidious in onset with multiple discrete lesions after release from treatment as a case of Hansen's disease seven years back. Initial FNAC report from a lesion revealed hemangioma which was unusual in presentation. He was later diagnosed clinically and histopathologically as a case of histoid leprosy.

Key words: Histoid leprosy, Lepromatous leprosy

Introduction

Histoid leprosy is an expression of multibacillary leprosy, characterized by typical cutaneous and subcutaneous nodules and plaques present over an apparently normal skin and histopathologically the predominance of spindle-shaped cells or polygonal cells and unusually large number of acid fast bacilli (AFB). The term histoid was originally coined in the year 1963 by Wade and Sehgal et al in the year 1988 documented the immunological profile of this unique expression of multibacillary leprosy. Histoid leprosy usually manifests itself on prolonged dapsone monotherapy or de novo without history of Hansen's disease. Histoid leprosy needs special attention in

post elimination era as emphasized by Sehgal et al (2009). There are few case reports on histoid leprosy after release from treatment, we need to study these cases better specially those with atypical presentation, characterize strain of *Mycobacterium leprae* from such cases for mutations and study for drug resistance for gaining in-depth understanding of etio-pathogenesis of histoid leprosy.

Case Report

A 64 years old male, farmer by occupation, presented with 03 months history of multiple reddish raised lesion over back, left thigh, chest and right leg. No history of pain, fever, swelling and weakness of hands and feet, difficulty in

Department of Dermatology Venereology & Leprology, Indian Naval Medicine Hospital Mumbai, 400005, Maharastra, India Corresponding author: Dr. Partha Sarathi Mohapatra e-mail: daringpartha@gmail.com

¹ Dr Partha Sarathi Mohapatra, MBBS, 3rd year Resident

² Dr Rahul Ray, MD Associate Professor

³ Dr Manish Khandare, MD, Senior Resident



Fig. 1: Non-tender nodules over Left thigh.



Fig 2: Non-tender nodules Right knee



Fig 3 : Non-tender nodules Right knee

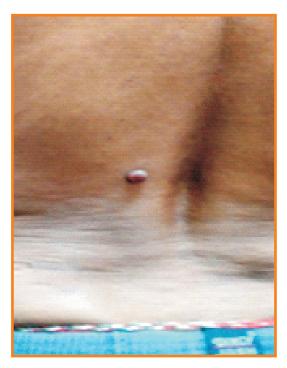


Fig 4 : Non-tender nodules on back

buttoning of shirt, slipping of chappals, stuffiness of nose, sensation of ants crawling under the skin and orchitis.

On past history patient was an old case of Hansen's disease, BT with type-1 reaction 7 years back and taken MB-MDT for one year along with tapering doses of corticosteroids. On cutaneous examination - few erythematous, non-tender nodules, firm in consistency, present over chest, left thigh (Fig 1), right above knee and knee (Figs 2, 3), back (Fig 4) and right lower 1/3rd of leg. Hypoaesthesia over both lower 1/3rd of leg as well as both feet and both hands predominantly over ulnar distribution was present.

On peripheral nerve examination - grade 1 thickness of right ulnar nerve, bilateral radial cutaneous nerve, right median nerve and right common peroneal nerve was observed. Grade-1 tenderness of right median and radial cutaneous nerve was also present. Egawa test on right hand was positive.

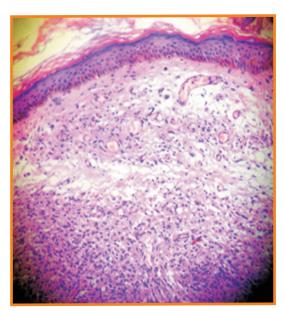


Fig. 5 : Foamy histiocytes filled with abundant Acid fast bacilli (H & E×100)

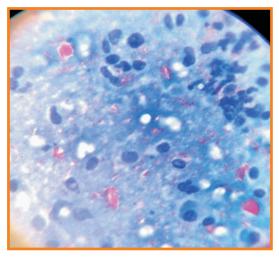


Fig. 6 : Collection of foamy histiocytes with Abundant Bacilli and Globi (Fite Faraco stain × 100)

Based on history and clinical examination provisional diagnosis of old case of Hansen's disease BT with multiple pilomatricoma with differentials of dermatofibroma or hemangioma was made. His all relevant hematological, biochemical and serological investigations were within normal limits. FNAC was performed from one of the lesions which was suggestive of hemangioma. Skin biopsy from the lesion confirmed it to histoid leprosy (Fig 5). On slit skin smear BI was 3.1 (Fig 6) and MI-1.3. Based on these histological and bacteriological findings this patient was diagnosed as a case of histoid leprosy and MDT was re-started again.

Discussion

Histoid leprosy is a well-known clinical entity usually occurring in patients on long-term diaminodiphenyl sulfone, with initial improvement followed by relapse. Irregular and inadequate therapies as well as resistance to dapsone and/or mutant organisms, may further compound its occurrence. A high degree of suspicion is

imperative in de novo cases (Bhat et al 2015). In addition to LL, histoid leprosy may additionally be seen in borderline and indeterminate leprosy groups also. Since the description of histoid leprosy by Wade, emphasis has been placed on retrospective histological diagnosis, which was subsequently reaffirmed (Sehgal et al 1988).

The term "histoid" was originally coined in 1963 by Wade for a bacillary-rich leproma composed of spindle-shaped cells, together with an absence of globus formation, so conspicuous in ordinary leproma, and exhibiting a fibromatoid tendency in chronic form (Wade 1963). Sehgal reported an incidence of 1.2% amongst LL patients (Sehgal and Srivastava 1988). The nodules of histoid leprosy may be soft, superficially placed, deeply fixed, or subcutaneous. Other lesions are neurofibroid (Meena et al 2017) or "molluscum-contagiosum-like" umbilicated papules (Ghorpade 2008). The number may vary from 3 to 50. The lesions are located on the back, buttock, face, extremities, or over bony prominences. Oral mucosal lesions are rarely seen. Palms, sole, and nasal mucosa are not affected commonly. Cutaneous nodules are firm, reddish or skin colored, dome shaped, oval or regular in contour, with shiny or stretched overlying skin. Cutaneous lesions may ulcerate at times. Subcutaneous nodules were commonly described by Wade earlier. The smaller nodules are soft and the larger ones are fibrotic. They may break down at the center. These lesions heal with scars. Cutaneous plaques develop on bony prominences which are sharply circumscribed.

Slit skin smears reveal abundance of organisms occurring either singly or in clusters known as "globi". The organisms appear as long rods with tapering ends, distinctly longer than the ordinary *lepra bacilli* (Sehgal and Joginder 1990). Histo-

pathology shows circumscribed nature of the lesion, the predominance of spindle-shaped cells and unusually large number of AFB. The circumscribed nature of the lesion is one of the cardinal features of histoid leprosy. It resembles a tissue forming process rather than a granuloma. Histoid lepromas, unlike ordinary lepromatous lesions, grow in an expansile, rather than infiltrative fashion. In most cases a free subepidermal zone is present. The lesion contains numerous, thin, spindle-shaped histiocytes forming interlacing bands, whorls, or tight curlicues (Job et al 1977). Our case report reaffirms the need of histopathology to confirm such cases, this becomes very important in cases with atypical presentation such as hemangioma in this case.

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