

A Case of Histoid Leprosy Presenting as Immune Reconstitution Inflammatory Syndrome (IRIS) in a Patient of Human Immunodeficiency Virus (HIV) Infection on Highly Active Retroviral Therapy (HAART)

Sir,
Histoid leprosy is a unique form of multibacillary leprosy with typical clinical morphology and histopathology. Leprosy patients co-infected with human immunodeficiency virus (HIV) infection have been described within the spectrum of leprosy as having either tuberculoid,^[1] borderline,^[1,2] or lepromatous^[1] leprosy. In our set up, so far HIV with leprosy infection have been noted in two cases which includes a borderline tuberculoid Hansen and the present case. Immune reconstitution inflammatory syndrome (IRIS) in HIV patients on highly active retroviral therapy (HAART) has been mentioned in the literature in cases with borderline tuberculoid^[3] or with type I reaction.^[4] Few case reports mention the co-infection of HIV and histoid leprosy.^[5,6] However, histoid leprosy manifesting as IRIS is hardly reported in the literature. Here we mention one such rare presentation.

A 51-year-old man, resident of Uttar Pradesh was diagnosed with HIV infection 1 year earlier and was on HAART (Tenofovir, Lamivudine, Efavirenz) for the last 11 months which he tolerated well. He was referred to our department for assessment of skin lesions when he had reported during a routine review at the ART centre. He complained of insidious onset of multiple asymptomatic raised skin-colored lesions over the chest, upper limbs, back, and face over the previous 7 months. There were no associated systemic complaints, numb patches, or shooting pain down the limbs. There was no history of contact with a known case of Hansen's disease.

General examination revealed a thin built individual with pallor. The systemic

examination was normal. Dermatological examination revealed the presence of multiple, polysized, discrete, normoesthetic, soft to firm, bilaterally symmetrical, dome-shaped skin-colored papules, plaque, and nodules over the chest, extensor aspect of both arms and forearms, back and left cheek [Figures 1 and 2]. There were no anesthetic patches or peripheral nerve thickening or signs of lepra reaction, deformity, or trophic ulcers. Differential diagnoses of sarcoidosis, leprosy, and lymphomatoid papulosis were made.

Further evaluation revealed hemoglobin of 8.6 g/dl, normal biochemical parameters, and a CD4 count of 126 cell/mm³. RNA load was found to be 20,000 copies/ml. Slit skin smear for acid-fast bacilli (AFB) showed bacteriological index (BI)-6+. Histopathology of a nodular lesion showed the presence of diffuse histiocytes and spindle-shaped cells [Figures 3 and 4]. Staining of tissue for acid-fast bacilli showed globular bacilli



Figure 1: Bilaterally symmetrical skin-colored papules, plaques, and nodules on the back

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Figure 2: Dome-shaped skin-colored nodules on the left cheek

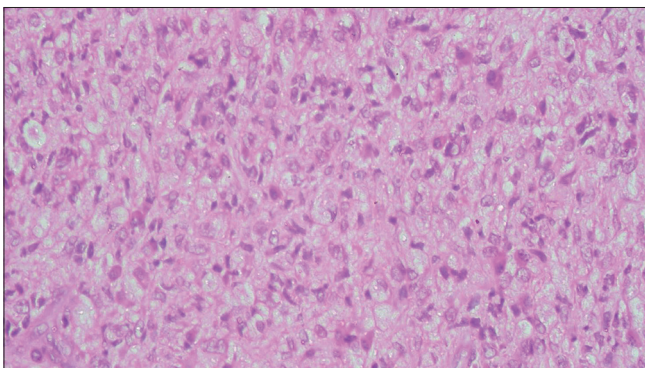


Figure 4: Histopathology showed diffuse spindle-shaped histiocytes (H and E 400x)

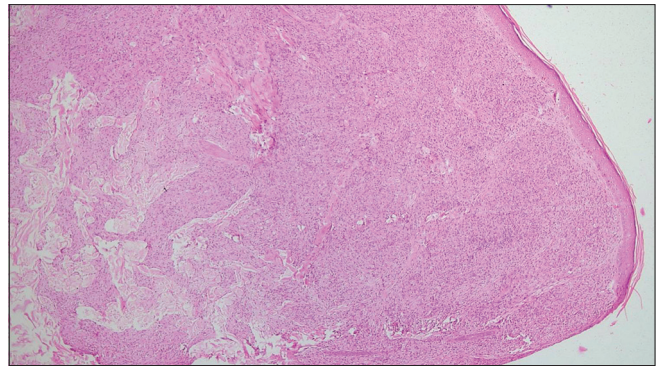


Figure 3: Skin biopsy from the nodular lesion showed diffuse spindle shaped histiocytes (H and E 100x)

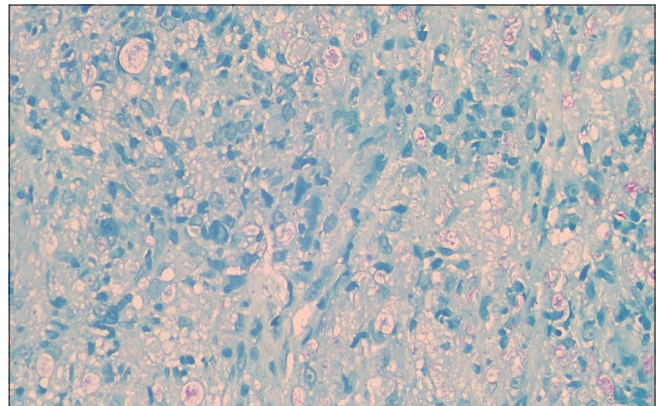


Figure 5: Ziehl Neelsen (ZN) stain (modified) of the tissue showed numerous AFB in clusters (oil immersion)

within the macrophages [Figure 5]. Based on the above findings, a diagnosis of Histoid leprosy in a case of HIV infection on HAART, with likely IRIS was made. He was not G6PD deficient. He was started on standard three-drug MDT (Rifampicin, Clofazimine, and Dapsone). In view of his anemia, he was managed with iron, folic acid, and multivitamin supplements. His hemoglobin and reticulocyte count were monitored periodically. There were no signs of hemolysis. He has completed 5 months of MDT so far and is responding well to treatment.

Histoid leprosy is clinically characterized by asymptomatic well-demarcated soft dome-shaped nodules and plaques appearing over apparently normal skin. The histopathology of lesions show elongated spindle-shaped macrophages with heavy bacillary load in the dermis and subcutis. The IRIS usually occurs between 4 and 6 months of starting HAART during which immune recovery is very rapid. The possible pathogenesis of leprosy IRIS is being studied from the various clinical presentations. It could be an inflammatory phenomenon which has unmasked the earlier undetected *Mycobacterium leprae* infection which has a long incubation period.^[7]

Case definition of IRIS associated leprosy, as suggested by Deps and Lockwood,^[8] is advanced HIV infection with (i)

a baseline low CD4+ count, (ii) presenting as Leprosy and/or reaction, and (iii) increase in CD4+ within 6 months of starting HAART. Similarly, Deps and Lockwood^[8] classified leprosy associated IRIS as four main types: type 1: unmasking – leprosy or type 1 reaction after starting HAART; type 2: overlap of immune restoration and leprosy diagnosed before starting HAART; type 3: undiagnosed leprosy occurring 6 months before HAART; type 4: unmasking followed by an overlap of immune restoration after HAART and MDT.

An association of histoid leprosy with HIV is quite rare.^[5,6] Bumb *et al.* reported Histoid leprosy in a HIV infected patient taking HAART for 9 months.^[5] Our case who was on HAART for the last 11 months, presented with asymptomatic skin lesions that developed 4 months after starting HAART. The patient had not taken medical assistance for nearly 7 months because the skin lesions were asymptomatic except for the cosmetic ailment. Over a period of 11 months, his CD4 count had increased from 56 to 126 cells/ μ l. Our case fits into the criteria of IRIS leprosy and in type 1 variant proposed by Lockwood, which is an unmasking of leprosy within 6 months of initiating HAART.

We present this rare and varied presentation of Hansen's Disease as in a highly endemic country like India,

Hansen's disease must always be kept in mind when any HIV-positive patient presents with varied skin lesions. The treating physician should be more vigilant in identifying such cases in the clinical setting of IRIS. Further research is required to understand the pathogenesis and pathology of this phenomenon of IRIS associated leprosy.

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Conflicts of interest

There are no conflicts of interest.

References

1. Pereira GA, Stefani MM, Araujo Filho JA, Souza LC, Stefani GP, Martelli CM. Human immunodeficiency virus type 1(HIV-1) and Mycobacterium leprae co-infection: HIV-1 subtypes and clinical, immunologic, and histopathologic profiles in a Brazilian cohort. *Am J Trop Med Hyg* 2004;71:679-84.
2. Rath N, Kar HK. Leprosy in HIV infection: Report of three cases. *Ind J Lepr* 2003;75:355-9.
3. Lawn SD, Wood C, Lockwood DN. Borderline tuberculoid leprosy: An immune reconstitution phenomenon in human immunodeficiency virus infected person. *Clin Infect Dis* 2003;36:5-6.
4. Kharkar V, Bhor UH, Mahajan S, Khopkar U. Type 1 lepra reaction presenting as immune reconstitution inflammatory syndrome. *Indian J Dermatol Venereol Leprol* 2007;73:253-6.
5. Bumb RA, Ghiya BC, Jakhar R, Prasad N. Histoid leprosy in an HIV positive patient taking cART. *Lepr Rev* 2010;81:221-3.
6. Kolaparambath BA, Rajagopal R. A case of histoid leprosy in a HIV infected person on HAART not responding to conventional MB MDT. *Indian J Lepr* 2014;86:165-9.
7. Geluk A. Correlates of immune exacerbations in leprosy. *Semin Immunol* 2018;39:111-8.
8. Daps P, Lockwood DN. Leprosy presenting as immune reconstitution inflammatory syndrome: Proposed definitions and classification. *Lepr Rev* 2010;81:59-68.