Borderline Tuberculoid Leprosy Associated with Histoid Leprosy

Angoori Gnaneshwar Rao

Abstract
Coexistence of two immunologically diverse forms of leprosy in an individual is very rare. Herein, we report a case of association of borderline tuberculoid (BT) leprosy with histoid leprosy (HL) in a young immune competent male. He was diagnosed as a case of BT leprosy 10 years ago and now presented with multiple papules and nodules. Histopathological examination of biopsy taken from patch and nodule showed features of BT and HL, respectively.

Key Words: Acid fast bacilli, borderline tuberculoid leprosy, histoid leprosy

Introduction
Leprosy is a chronic disease caused by Mycobacterium leprae affecting the peripheral nervous system, the skin, and other tissues. Histoid leprosy (HL) is an uncommon variant of lepromatous leprosy (LL), first described by Wade in 1960.[1] It is characterized by skin colored papules and/or subcutaneous nodules and plaques on apparently normal skin with distinct histopathology and characteristic bacterial morphology. It is usually seen in patients relapsing after dapsone monotherapy and also in the presence of dapsone resistance.

Case Report
A 27-year-old male presented with asymptomatic papules on the trunk and extremities of 5 months duration and not associated with epistaxis, ear lobe infiltration, glove and stocking anesthesia, or hoarseness of voice. No history of promiscuity or weight loss. He gives a history of the hypopigmented and hypoesthetic patch on the left leg 10-years ago, diagnosed as borderline tuberculoid (BT) leprosy and was advised for treatment. However, he developed severe drug reaction following intake of single tablet (most probably dapsone) which subsequently led to exfoliation of the skin. The episode of drug reaction desisted him from taking further treatment for leprosy. On examination, there was a single, ill-defined, dry, hypopigmented, and hypoesthetic patch on the medial aspect of the right knee [Figure 1]. Multiple dome shaped skin colored papules and nodules (total of 129 in number) ranging from 0.5 to 1.5 cm were distributed on the back, upper, and lower limbs. Lesions were not tendent, firm in consistency [Figure 2], and with intact sensations. Left ulnar and right lateral popliteal nerves were thickened and not tender. With these clinical features, he was offered a clinical diagnosis of BT leprosy with HL. Routine laboratory investigations were unremarkable. Slit skin smears from representative nodule and both ear lobes were positive for lepra bacilli that were both solid and granular and were long and slender [Figure 3]. Histopathological examination of biopsy from the patch (right knee) showed multiple epithelioid granulomas with few lymphocytes [Figure 4]. Biopsy from the papule showed spindle cells and foam cells arranged in interlacing bundles around adnexal structures and nerve twigs in the dermis [Figure 5], consistent with the diagnosis of BT leprosy with HL, respectively. He was started on antileprosy multidrug therapy consisting of rifampicin, ofloxacin, and clofazimine.

Discussion
The term histoid is derived from the microscopic appearance of the nodule showing spindle-shaped cells...
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resembling dermatofibroma. It is considered as a variant of LL by some researchers while others consider it as a distinct entity. An occurrence of 1.2% was reported among LL patients. The case under study is a young male aged 27 years, is in concert with reported male preponderance and the age group (21–40 years) reported in the literature. HL is known to be associated with dapsone resistance and also in relapse following dapsone monotherapy. However, it has also been reported with relapse in the presence of supervised monthly multidrug therapy and also in patients without any treatment. Histoid lesions rarely may occur along the course of peripheral nerve trunks and peripheral nerves. Morphology and distribution of skin lesions in the index case is in conformity with HL. There was no superciliary madarosis in the index case which is a notable feature in most of histoid patients and so is normal nasal mucosa. The cell-mediated immunity was found to be better in HL than in the active lepromatous patients as substantiated by increased CD36 expression by the keratinocytes, predominance of CD4 lymphocyte over CD8 lymphocytes, and increased number of activated lymphocytes and macrophages in the lesion. The humoral immunity was found to be enhanced as evidenced by increased absolute count of B lymphocytes and raised levels of immunoglobulins IgG, IgM, and IgA. Bacteriologically, there is large bacillary load in histoid lesions, which is attributed to focal loss of immunity. Furthermore, the acid-fast bacilli in these cases are measurably longer than ordinary lepra bacilli, are arranged in groups or parallel bundles in spindle-shaped histiocytes, described as histoid habitus by Wade. One of the significant electron microscopic features of histoid lesion is the absence of electron transparent zone or foam which is characteristically seen in lepromatous lesion. It is known that Electron transparent zone (ETS) interferes with bacterial metabolism leading to cell death. The absence of ETS is said to be responsible for the preservation of bacilli within the cells in histoid lesions.

The association of BT leprosy with HL in the index case is interesting as BT leprosy and HL belong to diverse
immunological spectrum, former being unstable and latter being stable form. In addition, it is intriguing to note how BT leprosy moved to HL (polar form of leprosy) if downgrading theory is contemplated. Furthermore, if BT leprosy is downgraded, then it should have moved to subpolar LL rather than to HL which is a polar form of leprosy. However, it is not known whether such association occurs due to unexplored underlying immunological mechanisms. On the contrary, the single hypoesthetic patch on right knee in the index case could not be considered as single lesion BL/LL, as the patch was dry and did not show shininess or infiltration and the histopathology also did not substantiate. Nonetheless, there have been reports of single lesion BL and LL cases reported in the literature albeit not in association with histoid lesions.[8,9] Review of literature could reveal only two case reports of association with HL; one with BT leprosy similar to the index case and the other with Indeterminate leprosy.[10,11]

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Conflicts of interest
There are no conflicts of interest.

References
1. Wade HW. The histoid leproma. Abstract. Int J Lepr 1960;28:469-70.
2. Price EW, Fitzhebert H. Histoid variety of lepromatous leprosy. Int J Lepr 1966;34:367-74.
3. Sehgal VN, Srivastava G. Histoid leprosy: A prospective diagnostic study in 38 patients. Dermatologica 1988;177:212-7.
4. Kalla G, Purohit S, Vyas MC. Histoid, a clinical variant of multibacillary leprosy: Report from so-called nonendemic areas. Int J Lepr Other Mycobact Dis 2000;68:267-71.
5. Weidmann MV, Argento CM, Garamuno R. Leprosy histioide of wade. Arch Argent Dermatol 2001;51:235-7.
6. Sehgal VN, Gautam RK, Srivastava G, Koranne RV, Beohar PC. Erythema nodosum leprosum (ENL) in histoid leprosy. Indian J Lepr 1985;57:346-9.
7. Job CK, Chacko CJ, Taylor PM. Electromicroscopic study of histoid leprosy with special reference to its histogenesis. Lepr India 1977;49:467-71.
8. Kar BR, Belliappa PR, Ebenezer G, Job CK. Single lesion borderline lepromatous leprosy. Int J Lepr 2004;72:45-7.
9. Yoder Lj, Jacobson RR, Job CK. A single skin lesion - An unusual presentation of lepromatous leprosy. Int J Lepr Other Mycobact Dis 1985;53:554-8.
10. Ramanujam K, Ramu G. Histoid transformation from unstable forms of leprosy. Abstract of Congress Papers 17/335. Int J Lepr 1963;41:685.
11. Das D, Gupta B, Deka B. De novo histoid leprosy with borderline tuberculoid leprosy: A rare association. J Evol Med Dent Sci 2014;24:6633-7.