Nodular Post Kala-Azar Dermal Leishmaniasis: A Report on Two Cases and Brief Review of Literature

Sir,

Post kala-azar dermal leishmaniasis (PKDL) occurs as a sequelae to visceral leishmaniasis and it is characterized by numerous hypopigmented macules, papulo-nodules, and plaques all over the body. The predominant lesions in immunocompetent individuals are macular and maculopapular, whereas nodular lesions are more common in immunocompromised patients. Besides, nodular lesions can be seen in long-standing cases of chronic PKDL.[1] We report two cases of nodular PKDL in the absence of any underlying immunosuppression.

Case 1

A 62-year-old male from Bihar presented with multiple nodules on face and extremities of 5 years duration. The lesions first appeared over face and lower limbs and then, gradually spread to the whole body. New lesions kept on appearing, whereas older lesions gradually increased in size. Lesions on right hand and scrotum got ulcerated spontaneously and healed slowly over the next couple of months. There was no history of sensory loss over lesions or extremities, joint pain, weight loss, or systemic symptoms. There was no history of prolonged fever associated with hepatosplenomegaly suggestive of kala-azar. On examination, multiple nontender, dusky erythematous papules, plaques, and nodules of various sizes were noted on the face and extremities [Figure 1a-d]. No sensory loss over lesions or extremities was demonstrable and peripheral nerve trunks were not thickened. Routine blood investigations, lipid profile, uric acid, rheumatoid factor, serology for human immunodeficiency virus (HIV), and X-ray of hands and feet were unremarkable.

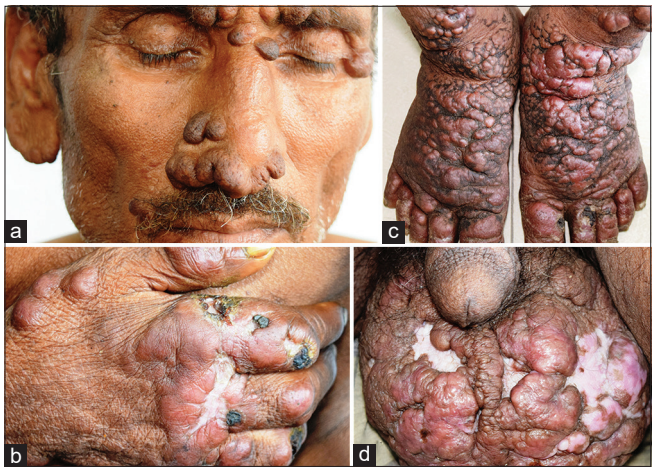

Figure 1: Multiple brown-red plaques and nodules on the face (a), right hand (b), bilateral feet (c) and scrotum (d). Note scars from healed ulcers over the hand and scrotum. (Case 1)

Case 2

A 40-year-old male from Bihar presented with multiple nodules around joints of extremities of 2 years duration and erythematous skin infiltration of face, trunk, and upper extremities of 1 year duration. Lesions first appeared over limbs and then, gradually spread to the whole body. New lesions kept on appearing, whereas older lesions gradually increased in size. However, none of the lesions ulcerated. There was no history of sensory loss over lesions or extremities, joint pain, weight loss, or systemic symptoms. There was no history of prolonged fever associated with hepatosplenomegaly suggestive of kala-azar. On examination, multiple nontender, juicy erythematous papules, plaques, and nodules of various sizes were noted on extremities, especially around joints [Figure 2a and b]. In addition, multiple hypopigmented macules and patches were noted on the trunk and upper extremities. Face including ear lobule was remarkable for diffuse erythematous infiltration. No sensory loss over lesions or

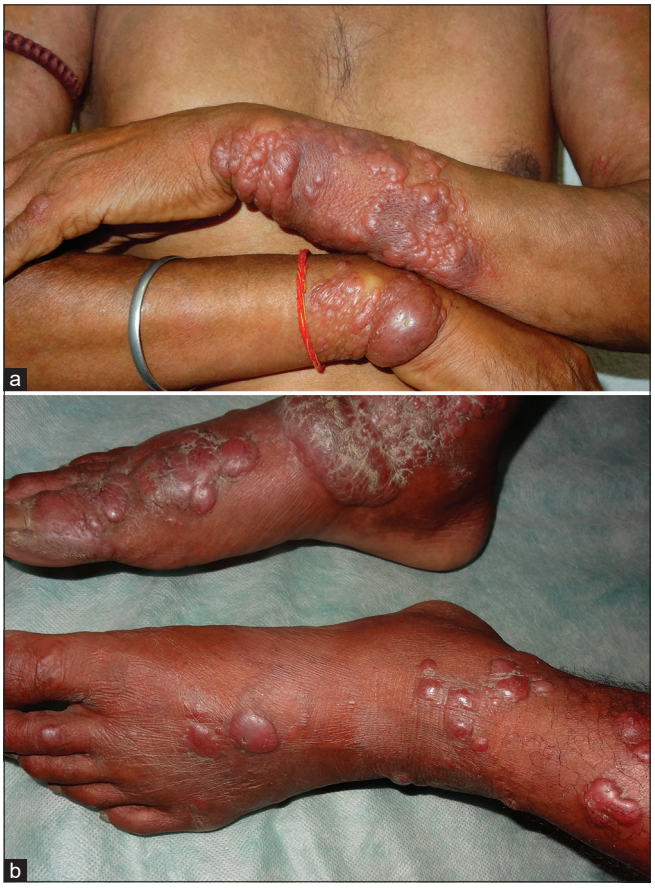

Figure 2: Juicy erythematous plaques and nodules on upper extremities (a) and feet (b). (Case 2)

extremities was demonstrable and peripheral nerve trunks were not thickened. Routine blood investigations, lipid profile, uric acid, rheumatoid factor, serology for HIV, and X-ray of foot were unremarkable. Slit skin smear from ear showed numerous Leishmania donovani (LD) bodies. Serology (rk39) was done and found to be positive.

In both the cases, histopathology showed atrophic epidermis, grenz zone, and curvilinear collection of macrophages, plasma cells, and some lymphocytes in upper dermis along with numerous LD bodies [Figure 3a-c]. Fite–Faraco stain did not show any acid-fast bacilli. Considering clinical and histopathological presentation, diagnosis of PKDL (nodular variant) was made. One patient was referred to government healthcare facility to receive amphotericin B under National Vector Borne Disease Control Programme (NVBCP), whereas the other patient was started on amphotericin B and he absconded after 2 injections.

PKDL was first described by Brahmachari in 1922. It usually occurs 6 months to 5 years after the treatment of visceral leishmaniasis, predominantly in the Indian subcontinent and Africa. In 15-20% of cases (consistent with our patients), there is no definite history of kala-azar but these patients hail from the endemic region in almost all the cases.[2]

Nodular lesions show a dense, diffuse dermal infiltrate of plenty of histiocytes and plasma cells. The infiltrate is characterized by a sharp margin, a feature which distinguishes the entity from lepromatous leprosy. Rarely, one may find diffuse infiltrate of foamy macrophages, mimicking leprosy. However, the presence of huge numbers of LD bodies provides a clincher for the diagnosis of PKDL.[3] Other histological findings include perineural infiltrates,[4] sclerosis around the dermal vessel walls.[5] Singh *et al*. had also reported the presence of atrophic epidermis and dermal adnexae being trapped in the dense infiltrate.[5]

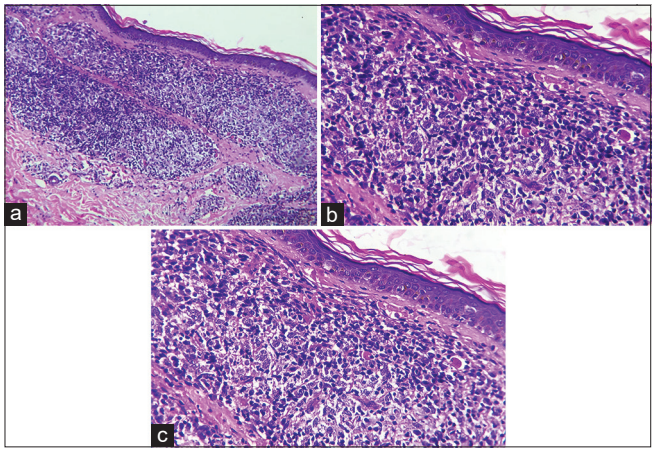

Figure 3: Thin atrophic epidermis, grenz zone, and curvilinear collection of macrophages, plasma cells, and some lymphocytes in the upper dermis, H and E x 100 (a) and Leishman donovan bodies, H and E x 400 (b). Close up of Leishman donovan bodies (c)

Clinical differentials for nodular PKDL include lepromatous and histoid leprosy, tuberous xanthoma, xanthoma disseminatum, cutaneous T cell lymphoma, etc. Besides, histological differentials for nodular PKDL include lymphocytomas, Jessner's lymphocytic infiltrate, pseudolymphoma, nodular lepromatous leprosy, histoplasmosis, etc.[5]

Therapeutic response is not satisfactory despite the availability of a wide number of drugs including pentavalent antimonials, amphotericin B, miltefosine, immunotherapy with Bacillus Calmette–Guérin (BCG) and heat-dead Leishmania amazonensis promastigotes. Combination therapy with liposomal amphotericin and miltefosine has been successful in a few cases. Poor cellular immunity makes this condition very difficult to treat and it is the sole cause responsible for the high rate of relapses in the course of the disease.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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