

Histoid Leprosy Presenting with Keloid Like Lesions

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A 42 year old male presented with multiple, discrete, hyperpigmented, firm, non elastic, non tender papules and plaques on the posterior trunk of 5 months duration, resembling keloid. The patient had also a few skin colored papules on the anterior trunk and face. The sensations over the skin lesions were intact. The patient had glove and stocking type of anesthesia and bilaterally thickened, non tender peripheral nerve trunks. The slit skin smear for acid fast bacilli from the ear lobes, skin lesions and normal skin were highly positive for *Mycobacterium leprae*. A skin biopsy showed a well defined collection of spindle shaped histiocytes in the dermis packed with acid fast bacilli. We are presenting here a case of histoid leprosy presenting with keloid like lesions, probably the rarest presentation of histoid leprosy.

Key words : Histoid leprosy, Keloid, Atypical presentations

Introduction

Histoid leprosy (HL), a rare variant of lepromatous leprosy (LL), first described by Wade, is a type of leprosy first described in patients who were on dapsone monotherapy, irregular therapy and inadequate therapy, indicating drug resistance (Wade 1963). However, *de novo* cases are being increasingly reported (Nair and Kumar 2013, Sehgal et al 2009). HL presents with localized skin lesions arising from an apparently normal skin, in contrast to LL where the lesions arise from an infiltrated skin. Such unusual presentation along with lack of infiltration of the ear lobes and intact sensations over the skin lesions may become a cause for misdiagnosis. Moreover HL can present

with a myriad of skin lesions mimicking many dermatoses. We are reporting here a case of HL presenting with keloid like lesions.

Case Report

A 42 year old male presented with asymptomatic skin lesions of 5 months duration. The lesions started as a few papules on the posterior trunk below the left scapula and over a period of 5 months, similar lesions appeared on other areas of the posterior trunk. The patient had also noticed the appearance of a few skin colored papules on the anterior trunk and face. The patient did not have any constitutional symptoms and denied history of trauma at the site of the skin lesions. The patient did not give any past history

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Fig 1 : Close view of keloid like papules and plaques on the posterior trunk

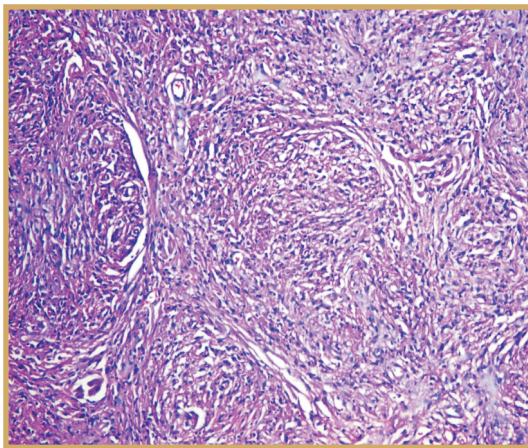


Fig 2 : Spindle shaped histiocytes in the dermis arranged in whorls and interwining crisscross pattern, H&E x400

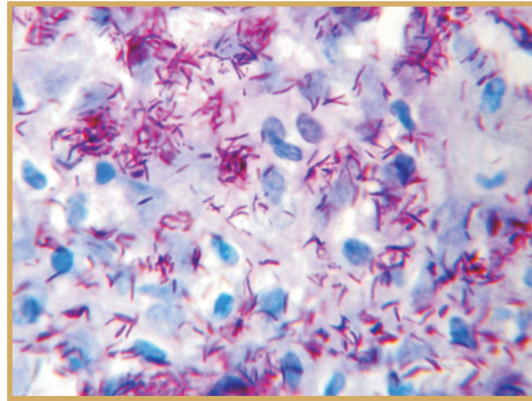


Fig 3 : Histiocytes packed with acid fast bacilli which are long, narrow with thin tapering ends, Wade-Fite stain x1000

of transient tender erythematous subcutaneous nodules.

On examination the patient had multiple, discrete, firm, rough, hyperpigmented, non tender, papules and plaques distributed on the posterior trunk in a localized and asymmetrical pattern, arising from an apparently normal skin and resembling keloids (Fig 1). The patient also had a few soft, skin colored, papules on the anterior trunk and face, arising from non infiltrated skin. There was no ear lobe infiltration or superciliary madarosis. The sensations of temperature, touch and pain were intact over the skin lesions. The patient had glove and stocking type of anesthesia. The ulnar, common peroneal, superficial peroneal and posterior tibial nerves were bilaterally thickened, but non tender. The patient did not have any motor deficit or disability or deformity. His systemic examination was unremarkable.

The patient's blood hemogram, biochemistry, liver and renal function tests, chest x-ray and ultrasound abdomen were within normal limits. A slit skin smear (SSS) for acid fast bacilli (AFB) from the ear lobes, keloidal papules, skin colored

Table 1 : Review of the atypical presentations of histoid leprosy

No.	Author	Year	Atypical presentations
1	Rodriguez JN.	1969	Papules and nodules on the hard palate and glans penis
2	Chaudhary DS et al	1971	Papules and nodules with ulceration on the trunk and extremities
3	Girdhar A et al	1990	Nodules along peripheral nerve trunks resembling nerve abscess
4	Baslas RG et al	1992	Papules and nodules on the palms
5	Rajan MA	1998	Nodule on the episclera
6	Thappa DM et al	2001	Nodules resembling tuberose xanthomas on the elbows, knees and ankle
7	Shivaswamy KN et al	2004	Multiple sinuses on the scrotum
8	Nair SP et al	2006	Giant pedunculated tumor like masses on the right thigh resembling neurofibroma, dermatofibroma
9	Ghorpade AK	2008	Molluscum contagiosum like papules with pseudo-Koebner phenomenon on trunks and limbs
10	Ghorpade AK	2010	Eroded papules and nodules resembling trans-epidermal elimination on face and extremities
11	Present study	2014	Keloid like papules on the posterior trunk

papules and normal skin, stained by Ziehl Neelsen technique, were positive for *Mycobacterium leprae*, with a bacterial index of 6+ (>1000 bacilli/oil immersion field) and the morphological index of 40% from all sites. A skin biopsy from both the keloidal and skin colored papules and stained with hemotoxylin and eosin showed identical findings. The dermis showed a well defined collection of spindle shaped histiocytes arranged in whorls with interwining, crisscross pattern (Fig 2) and Wade-Fite stain demonstrated the histiocytes to be packed with AFB (Fig 3). We made a final diagnosis of HL presenting with keloid like lesions and the patient was started on WHO multibacillary multidrug therapy (MB-MDT), comprising daily dapsone 100 mg, clofazimine 50 mg and monthly pulses of rifampicin 600 mg and clofazimine 300 mg. We also added ofloxacin 400 mg daily.

Discussion

The presence of skin colored papules on the trunk and face, rising from normal skin, glove and

stocking type of anesthesia and nerve thickening prompted us to suspect HL. However we were confounded by the keloidal papules on the posterior trunk. Keloid is known to occur following trauma, *de novo* and also following an episode of necrotic erythema nodosum leprosum (ENL). Our patient did not give any history of trauma, nor did he have a past or present clinical evidence of ENL. The SSS from the keloidal papules were positive for AFB and the histopathology showed the classical features of HL and no features of keloid. Therefore we made a diagnosis of HL presenting with keloid like lesions.

HL classically presents with smooth, shiny, dome shaped, soft to firm papules, nodules and plaques, in contrast to the hyperpigmented, firm, rough, papules and plaques on the posterior trunk of our patient which resembled keloid. HL can present with protean manifestations (Baslas et al 1992, Chaudhury et al 1971, Girdhar et al 1990, Rajan 1998, Rodriguez 1969, Shivaswamy et al 2004, Thappa et al 2001), such uncommon

presentations may confuse even astute and highly experienced clinicians. The skin lesions vary from molluscoid lesions (Ghorpade 2008) to giant tumor like masses (Nair et al 2006). Some of atypical manifestations of HL are listed in Table 1. HL is raising concerns among leprosy workers, as they are highly bacilliferous, can be missed clinically and is a potential reservoir of mutant drug resistant bacilli (Nair and Kumar 2013). A recent study has demonstrated that both the spindle shaped histiocytes of HL and the foamy macrophages of LL are CD68 positive, indicating common macrophage lineage (Da costa et al 2013). The presence of epidermal atrophy, grenz zone and spindle shaped histiocytes arranged in whorls, circles, with intertwining crisscross patterns in the dermis, surrounded by a pseudocapsule and packed with AFB are the histopathological hallmarks of HL (Wade 1963, Nair and Kumar 2013, Sehgal et al 2009). The *M leprae* in HL lack globi because of the absence of a matrix secretion "gloea" and the bacilli are long, narrow with thin tapering ends (Fig 3), indicating them to be mutant strains (Nair and Kumar 2013). The bacilli in HL are mainly solid forms tightly packed within the histiocytes along the long axis of the cells without distorting the cells and this is denoted as the "histoid habitus", in contrast to LL where the bacilli includes fragmented and solid forms, distorting the foamy macrophages (Nair and Kumar 2013, Sehgal et al 2009).

HL usually responds to the conventional MB-MDT, but addition of ofloxacin (Vora et al 1995) or *Mycobacteria w* (Talwar 1999) vaccine will have the potential to reduce the bacillary load faster. *Mycobacterium w*, renamed as *Mycobacterium indicus pranii* (MIP) is among effective immunomodulators that has shown beneficial immuno-

therapeutic effects in leprosy in several studies (Katoch 2016). As histoid leprosy may have role in transmission (Ghorpade 2010), such adjunct therapy may also be helpful in reducing the chances of such transmission. Keloidal presentation has been described in a case of LL (Yoder et al 1985), but not in HL to the best of our knowledge. We are presenting here with one more atypical presentation of HL as keloid.

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