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Case Report

Lepromatous leprosy: An Unusual Presentation

SK Gupta[1], S Kaur[1], V Malhotra[1], AK Arora[1], N Sood[2] and V Gupta[3]

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A 25 year old man presented with loss of sensations over both hands and feet and extreme difficulty in passing urine. On examination, an indurated sclerotic plaque was present on shaft of penis and scrotum in addition to other features of lepromatous leprosy. Skin biopsy from the penile lesion showed presence of a large number of acid fast bacilli with a BI of 6+ extending into the epidermis and histopathology showed absence of Grenz zone and presence of foamy macrophages in the dermis upto the dermo-epidermal junction.

Key words : Lepromatous leprosy, penile lesions, Grenz zone

Introduction

Leprosy is caused by *Mycobacterium Leprae*, which has a distinct predilection for cooler parts of body (Anish 1971). Only a few cases of genital involvement in leprosy have been reported so far (Kumar et al 2001). Though scrotal involvement is observed in most of the cases of genital involvement in leprosy reported till date, involvement of the penile shaft has been described rarely (Shaw et al 2002). Present case is being reported because of very rare involvement of penile skin in form of a sclerotic plaque resulting in shortening and partial amputation of penis.

Case Report

A 25 year old unmarried man, permanent resident of a leprosy endemic area, presented with loss of sensation over hands and feet, with resultant multiple ulcers, fissures and inability to do routine work, for the last 5 years. He also complained of thickening and tightening of skin over the external genitalia with difficulty in passage of urine for the last three years. The patient gave history of chronic nasal blockage along with episodes of epistaxis, and recurrent swelling of hands and feet which had now become permanent for the last seven years. For the last five years there was loss of sweating

[1] SK Gupta, MD, Prof & Head, Department of Skin & VD, Dayanand Medical College and Hospital, Ludhiana.
[1] S Kaur, MD, Department of Skin & VD, Dayanand Medical College and Hospital, Ludhiana.
[1] V Malhotra, MD, Department of Skin & VD, Dayanand Medical College and Hospital, Ludhiana.
[1] AK Arora, MBBS, Department of Skin & VD, Dayanand Medical College and Hospital, Ludhiana.
[2] N Sood, MD, Department of Pathology, Dayanand Medical College and Hospital, Ludhiana, India.
[3] V Gupta, MD, Department of Microbiology, Dayanand Medical College and Hospital, Ludhiana, India.
Correspondence to: SK Gupta **Email:** vsunilgupta@rediffmail.com

over the extremities and excessive sweating on face, trunk and axilla. The patient also complained of a visible ear and nose shape deformity which had been present for the last seven and four months respectively. There was no history of recurrent development of erythematous tender nodules on the body or constitutional features. The patient was taking some indigenous medication for the above complaints same, off and on, but with no relief. The patient did not give any history suggestive of intake anti leprosy Multi Drug Therapy (MDT).

Cutaneous examination showed diffuse infiltration and nodularity on the face. There was loss of eyebrows, eyelashes, and moustache at places with absence of beard. There was thickening of ear lobes with loss of upper part of the right ear cartilage and lateral part of the left ear cartilage. Saddle nose deformity was seen with collapse of the bridge of nose. (Fig 1) Thickening of the lips and nodules over the tongue and palate and buccal mucosa were observed. Diffuse symmetrical infiltration of the skin over the trunk, buttocks and extremities could be appreciated. On the trunk symmetrical small hypopigmented macules could also be appreciated around both the flanks. Absence of hair in axillae, pubic area and limbs was observed. Swelling of both hands and feet was seen along with clawing of the little fingers of both hands, and with multiple ulcers and fissures on both feet.

The examination of the genitals revealed gross deformity of the external genitalia. Erythematous indurated sclerotic plaque could be appreciated on the prepuce and skin over the penile shaft and anterior two-thirds of scrotum, as a result of which the penile shaft appeared much smaller, with loss of the prepucial skin and the urethral meatus was stenosed. (Fig 2) Rugosities over the scrotum were lost. On questioning, the patient also gave history of erectile dysfunction which could be inferred from the amputation and shortening of the penis.

Sensory examination revealed no loss of sensation over the body or skin lesions apart from hands and feet where hypoesthesia was present in a glove and stocking appearance, and there was

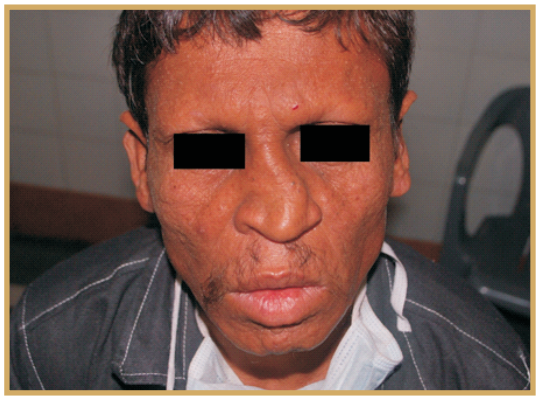

Fig 1 : Diffuse infilteration of face with development of nodules at places along with madarosis and sadlle nose deformity.

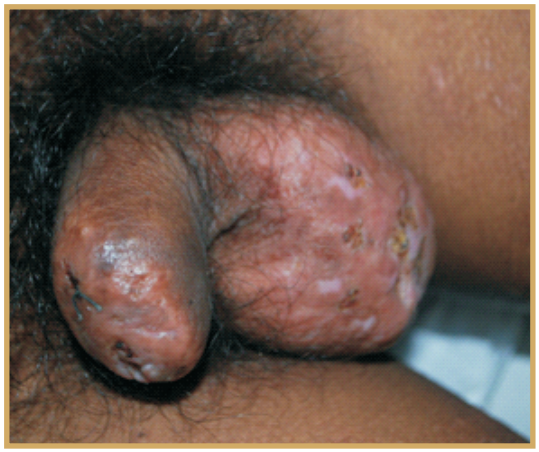

Fig 2 : Thick indurated sclerotic plaques could be appreciated on the penile shaft and prepuce resulting in auto amputation of penis and phimosis.

loss of testicular sensation. Peripheral nerve examination showed bilaterally symmetrical enlargement of ulnar, radial cutaneous, greater auricular, lateral popliteal and posterior tibial nerves. The nerves were non tender, smooth and firm.

Complete hemogram, liver and renal function tests, and urine analysis were normal. The slit skin smear examination stained by modified Ziehl-Neelson's (ZN) staining method showed presence of globi with a BI of 6+. Histopathologic examination of skin biopsy from the trunk revealed an atrophic and flattened epidermis along with a dense collection of foamy macrophages in the dermis and a clear subepidermal "Grenzzone". Modified (ZN) stained tissue section showed an abundance of AFB visible as globi with a BI of 6+ (Fig 3a,b). However, histopathologic findings of skin biopsy from the penile plaque showed a diffuse collection of foamy macrophages without any free subepidermal Grenz zone. On modified ZN staining, AFB were seen in clumps in the dermis and were also observed lying intracellularly, both singly and in clumps, throughout the layers of the epidermis giving rise to a BI of 6+ (Fig 4a,b,c). There was no difference in the BI calculated from either slit skin smear examination or biopsy examination. Patient was prescribed WHO multidrug therapy, multibacillary regimen, along with supportive care for the plantar ulcers. As the patient suffered from difficulty in passing urine, he was prescribed tab urimax 0.4 mg twice daily and urethroplasty was planned in future in case of no relief with medical treatment. However he returned to his home state and was lost to follow up.

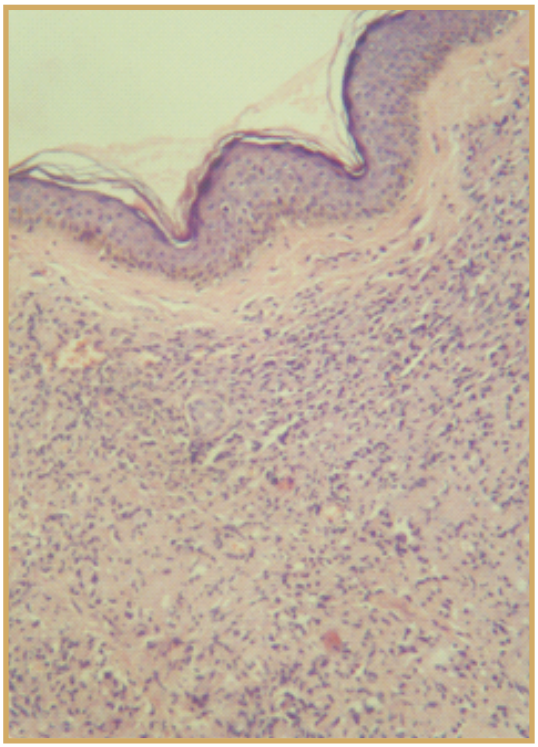

Fig 3a : Photomicrograph (H&E X100) of skin biopsy from trunk showing epidermal atrophy with loss of rete ridges, presence of subepidermal grenz zone and diffuse foamy macrophage granuloma in the dermis.

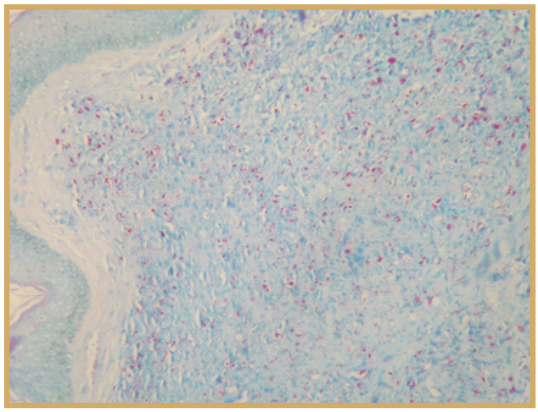

Fig 3b : Photomicrograph (Modified ZN stain X 200) of truncal biopsy showing presence of solid staining AFB singly and as globi inside macrophages in the dermis.

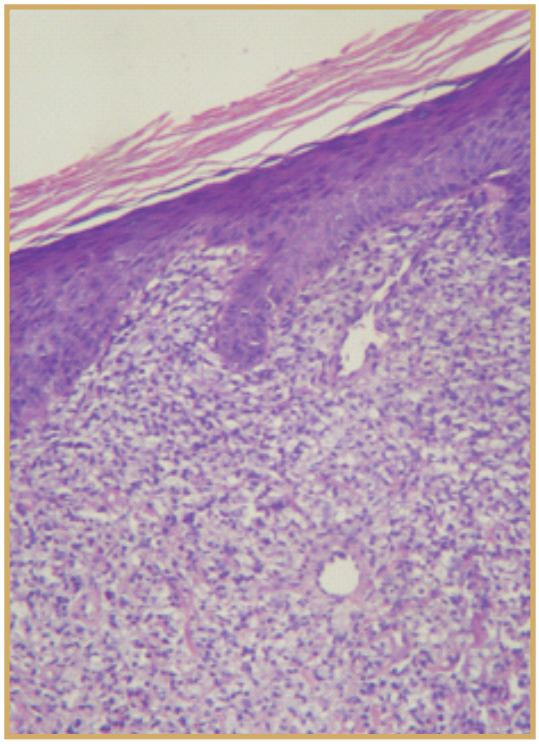

Fig 4a : Photomicrograph (H&E X 100) of penile biopsy showing foamy macrophages infiltrating the entire dermis with absence of grenz zone.

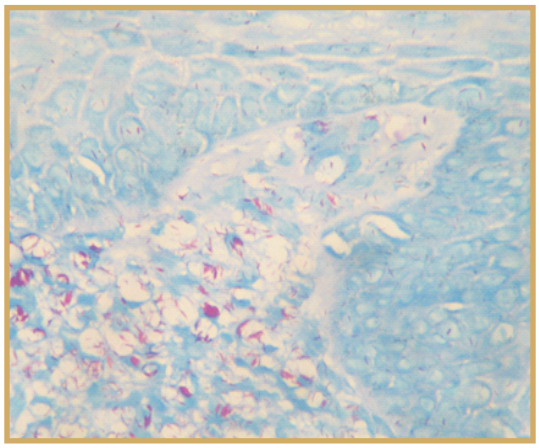

Fig 4b : Photomicrograph (Modified ZN stain X 800) of penile biopsy showing presence of solid staining AFB the invading the epidermis.

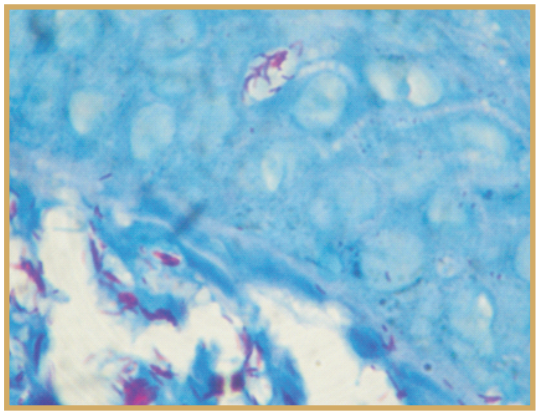

Fig 4c : Modified ZN stainining of penile biopsy showing presence of AFB present as globi within a vacuole inside keratinocyte. (X 2000)

Discussion

Lepromatous leprosy is the most severe form of leprosy as it can involve almost any part of the human body. However, certain zones like scalp, palms and soles, genitalia, groins, axillae, eyelids, a transverse band of skin over lumbosacral area, midline of back and perineum have been defined as being immune to the development of leprosy attributed to the relatively high local temperature of these sites. (Rajashekhar et al 2009). However, though infrequently, involvement of these sites in leprosy has been reported.

Under experimental conditions, scrotal skin has been reported to be cooler than the core body temperature, (Kandeel and Swerdloff 1998) and leprous granulomas and AFB have been found in one - third of biopsies of patients (treated and untreated) with all types of leprosy even in the absence of lesions on the scrotum. (Pandya and Antia 1974). With the use of occlusive undergarments, it is likely that the temperature of the penile and scrotal skin may remain elevated making the area immune or less prone to

the development of leprosy (Kumar et al 2001). However, in India where people are more used to wear loose underclothes, we are likely to encounter more cases of genital leprosy.

Genital leprosy has been reported previously in all types of the disease spectrum (Rajashekhar et al 2009, Kumar et al 2001). Primary involvement of scrotum has been reported in the indeterminate (Murthy et al 1993) and tuberculoid type (Dixit et al 1990) of leprosy. Arora et al (1989) found cutaneous lesions of male genitalia in 2.9% cases examined and most of them were of borderline type. Clinical involvement of scrotal skin in lepromatous leprosy in the form of nodules and hydrocele has also been described in fresh as well as relapsed cases of lepromatous leprosy and histoid leprosy. (Ebenso 2000, Thappa et al 1999, Nigam and Singh 1990, Ramanujan and Ramu 1969) Kumar et al (2001), observed 6.6% cases with genital lesions of 467 male leprosy patients in one study, where genital lesions were most frequently encountered in LL (25.8%) followed by BL (13.3%) and BT (1.4%) leprosy.

However, to the best of our knowledge, cases as mutilating as the present case, resulting in partial auto amputation of penis and severe phimosis, have never been reported in lepromatous leprosy. The extent of involvement was so severe that it led extreme difficulty in passing urine.

Another interesting feature pertaining to this case is the difference in the Haematoxylin and Eosin (H& E) stained sections of both the biopsies in this patient. The absence of Grenz zone in the penile biopsy and presence of AFB in the epidermal cells is rather unusual. It is possible to find bacilli as artefacts in epidermis as occasionally AFB attached to microtome knife can be transferred to other areas of section. These artefacts are known as “floaters” (Satapathy et al 2005). However, in the present case it can be recognized that bacilli were arranged inside the vacuoles of cytoplasm of keratinocytes and therefore were not floaters. (Fig 4a, 4b,4c)

The H&E and Harada's modified Allochrome method for AFB revealed presence of AFB in epidermal cells of apparently normal skin of all forms of leprosy but AFB were significantly low in inderteminate and TT forms of leprosy than in the BL and LL forms (Budhiraja et al 2012). The presence of bacilli in keratinocytes can be due to phagocytic activity of keratinocytes which engulf bacilli from subepidermal zone. The presence of AFB in epidermis strongly suggests that dissemination of leprosy bacilli can occur from skin to skin contact.

Unfortunately, as the patient was lost to follow up we failed in our ultimate aim of curing the patient, helping him out of his disabilities, and documenting his treatment response which is also a shortcoming of the present case.

Thus, though infrequent, genital involvement and presence of bacilli in epidermis of lepromatous skin biopsies, has been reported in leprosy. Therefore, genital examination should be done in all patients presenting throughout the spectrum of leprosy to prevent additional morbidity in the patients. Also, the possibility of skin as a route of transmission of leprosy should be seriously considered.

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