

Histoid leprosy on scrotal skin: an unusual case

Paudyal P,^{1*} Shrestha S,¹ Agarwal M,¹ Kafle SU,¹ Karki S,¹ Upadhyaya P,¹ Agarwal S,² Pradhan A¹

¹Department of Pathology, B.P. Koirala Institute of Health Sciences, Dharan, Nepal; ²Department of Dermatology and Venerology, B.P. Koirala Institute of Health Sciences, Dharan, Nepal

*Corresponding to: Dr. Punam Paudyal, Department of Pathology, B. P. Koirala Institute of Health Sciences, Dharan, Nepal, email: punamzpranju@hotmail.com, Tel No.: (+977) 9842040269

ABSTRACT

INTRODUCTION: Although no part of the skin is immune from invasion by mycobacterium leprae it is commonly seen over cooler parts of the body and very rarely found over external genitalia because of their warm temperature. Histoid leprosy is rare but a well-defined entity with specific clinical, histopathological, and bacteriological features.

CASE REPORT: A Punch biopsy measuring 3x3 cm of a 73 year old male having an erythematous plaque over scrotum since 2 months was received for histopathologic examination with the clinical differentials of pilomatrixoma and steatocystoma multiplex. Histopathologic and special stain slides revealed a feature of histoid leprosy.

CONCLUSION: Although rare, leprosy lesions may occur on the male genitalia and therefore in all male patients, history taking and examination of external genitalia should not be neglected.

KEY WORDS: Histoid leprosy, Scrotal skin, Bacillary index

Article submitted 28 March. Reviewed 20 April. Author correction 25 April. Final version accepted 27 May 2013.

INTRODUCTION

Hansen's disease is a chronic infectious disease caused by *M. leprae*, affecting the peripheral nerves, skin, and certain other tissues which are found predominantly in cooler parts of the body and are more exposed to trauma.^{1, 2} Although involvement of the male genitalia, particularly of the gonads, is well known in leprosy, lesions of leprosy are not commonly found on the genital skin and there have been only a few reports of such involvement.³

Histoid leprosy is an uncommon variant of lepromatous leprosy with characteristic clinical, histopathological and bacteriological findings. It was originally described by Wade in 1963 as discrete, firm lesions and dome-shaped nodules which develop on apparently normal skin in patients with lepromatous leprosy. The pathogenesis of this rare and unusual variant of leprosy remains unresolved. The interplay of genetic factors, immune response and treatment received seems to influence the manifestations of Histoid leprosy.⁴

Here we report a case of histoid leprosy considering its rarity, as well as its variable clinical expression and uncertainty regarding its etiopathogenesis which clinically manifested as a cutaneous lesion in the scrotal skin with positive acid fast bacilli (AFB) in Ziehl Neelsen (ZN) stain.

CASE REPORT

A 73 year old male presented to the outpatient department of dermatology and venerology with chief complaints of redness over scrotum for 2 months. Lesion was gradually progressive, associated with occasional itching. Examination of the lesion revealed an erythematous plaque of size 3x3 cm, irregular in shape with well defined margin and central pallor. The surface was smooth, firm to hard in consistency with absence of tenderness and normal overlying temperature. There was neither history nor clinical evidence of other hypopigmented, hypoanesthetic skin lesions, motor weakness, or glove-and-stocking anesthesia. There was no history of any drug intake. All the hematological and biochemical investigation were within normal limits. Based on the cutaneous lesion on the scrotal skin pilomatrixoma and steatocystoma multiplex were considered as provisional diagnosis. Punch biopsy of the lesion measuring 3 x 3 cm was sent for the histopathological evaluation. On histopathological examination the dermis showed proliferation of foamy histiocytes along with interlacing

bundles of spindle shaped cells in storiform pattern involving the entire dermis (Figure 1). These cells were also seen involving the nerve bundles (Figure 2) and erector pili muscles.

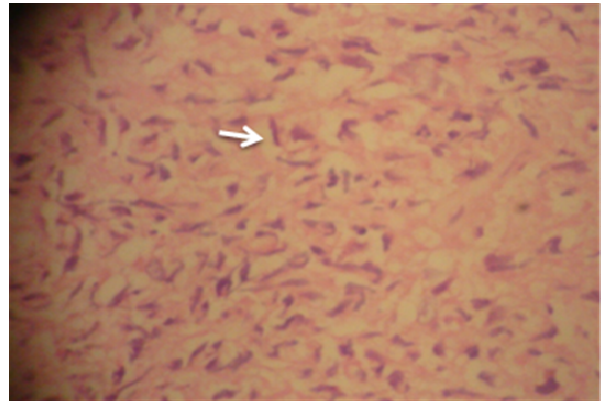


Figure 1. Foamy histiocytes admixed with spindle shaped cells (shown by arrow) arranged in storiform pattern (20 X, H&E)

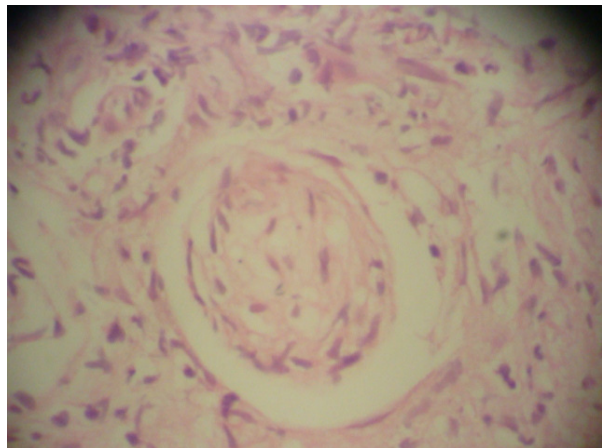


Figure 2. Perineural involvement by the foamy histiocytes and spindle shaped cells (40 X, H&E)

Keeping the possibility of leprosy in mind, ZN stain for AFB Leprosy was done. The ZN stain slide revealed abundant positive acid fast bacilli for leprosy.

According to the Ridley's logarithmic scale the bacillary index (BI) was 6, i.e. >1,000 bacilli per high power field (Figure 3). Based on the histomorphologic and special stain findings, the diagnosis of histoid leprosy was made.

DISCUSSION

The scrotal skin has been described as an unusual site for leprosy, although no part of the skin is immune from invasion by *M. leprae*.^{5,6} The scrotal

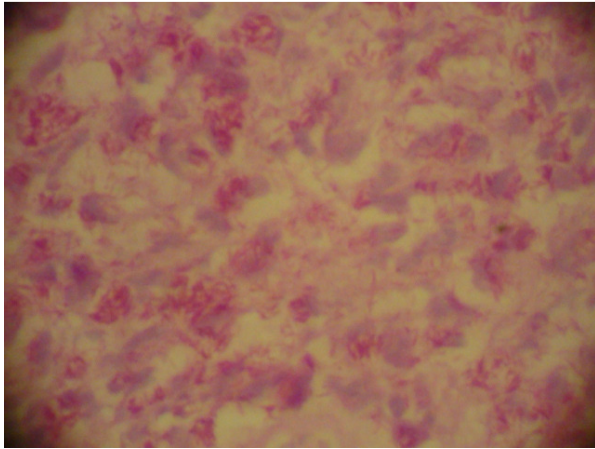


Figure 3. Abundant long slender acid fast bacilli laden cells with the bacillary index of 6 (100 X, ZN)

region being relatively cooler than the core body temperature is expected to be vulnerable for infiltration with *M. leprae*. However, because of the use of tight undergarments, the temperature of the scrotal skin is likely to be elevated making this area less prone to the development of leprosy lesions.^{3,5} The disease is endemic in many tropical and subtropical countries but is declining in prevalence as a result of multidrug therapy. The Indian subcontinent, Southeast Asia, sub-Saharan countries in Africa and Brazil comprise the areas most affected at present.^{7,8}

Genital lesions have been mostly described in adults, the youngest being a 12-year-old boy.⁹ In children, it presents predominantly as paucibacillary disease, and there are only a few reports of genital involvement.^{9,10}

Parikh et al reported six cases with leprosy lesions of the penis and scrotum.¹⁰ Histopathological and bacteriological involvement of the scrotal skin in lepromatous leprosy has been documented by Pandya et al and Ramu et al.^{11,12} Pandya and Antia reported leprosy granulomas and AFB in one third of biopsies of scrotal skin in leprosy patients even in absence of lesions in the scrotum.¹¹ In a study by Ramu et al scrotal biopsies were obtained from 38 cases of lepromatous leprosy who had clinically subsided lesions with negative skin smears. Twenty-six (68.4%) of these cases revealed bacilli in the dartos muscle.¹² Arora et al found leprosy lesions on external genitalia in 13 (2.9%) patients after examining 450 male leprosy patients. In all 13 cases the scrotum was involved.¹

Histoid leprosy has been rarely reported in genital lesions.^{4,6,12}

The term Histoid leprosy was first coined by Wade in 1960 as a histological concept of bacillary rich leproma composed of spindle shaped cells along with an absence of globus formation. Since then few case series have been published mostly from India. Histoid leprosy occurs in lepromatous patients who relapse after dapsone monotherapy, in those with dapsone resistance, sometimes even after multidrug treatment, or at times de novo.⁴

Though, presentation of leprosy has been shown to decrease at the primary health care level, due to its near elimination; multibacillated forms will still continue to present in our speciality especially at a tertiary care level.¹³

This case point out that, there is gross disparity in the clinical diagnosis and histopathological diagnosis, which always remains a reliable diagnostic tool.

CONCLUSION

This report emphasis the need for a thorough genital examination in all male patients and histopathological evaluation of all suspected lesion in scrotum, so that early anti-leprosy treatment can be initiated and subsequent complications of impotence and sterility can be prevented. This case highlights that Histoid leprosy should be kept in mind in the differential diagnosis of genital lesions.

CONFLICT OF INTEREST: None to declare.

FINANCIAL INTEREST: None to declare.

REFERENCES

1. Arora SK, Mukhija RD, Mohan L, Gridhar M. A study of cutaneous lesions on male external genitalia. *Indian J Lepr* 1989;61:222-224.
2. Anish SA. The relationship between surface temperature and dermal invasion in lepro- matous leprosy. *Int J Lepr Other Mycobact Dis* 1971; 39:848-51.
3. Fox H, Knott J. Leprous nodules on male genitalia. *Int J Lepr* 1934;2: 445-446.
4. Wade HW. The histoid variety of lepromatous leprosy. *Int J Lepr* 1963;31;129-43.
5. Cochrane RG, Davey TF. Ulnar and lateral popliteal nerve involvement in Relation to low temperature zones. In: *Leprosy in theory and practice*. 2nd ed. Bristol: John Wright and Sons Ltd; 1964. p. 266.

Leprosy of scrotal skin

6. Kaur S, Kumar B. Study of apparently uninvolved skin in leprosy as regards bacillary population at various sites. *Lepr Ind* 1978;50:38-44.
7. Kumar B, Kaur I, Rai R, Mandaal SK, Sharma VK. Involvement of male genitalia in leprosy. *Lepr Rev* 2001;72:70-77.
8. Noordeen SK. Eliminating leprosy as a public health problem. *Int J Lepr* 1995;63 :559.
9. Sehgal VN, Koranne RV, Sharma AK et al. Age at onset of leprosy. *Lepr Ind*, 1982; 54: 332-337
10. Parikh DA, Parikh AC, Ganapati R. Penile and scrotal lesions in leprosy : case reports. *Lepr Rev* 1989;60:303-305.
11. Pandya NJ, Antia NH. The value of scrotal biopsy in leprosy. *Lepr Rev* 1974;45:145-152.
12. Ramu G, Desikan KV. A study of scrotal biopsy in subsided cases of lepromatous leprosy. *Lepr India* 1979;51:341-347.
13. Mendiratta V, Jain A, Chander R, Khan A, Barara M. A nine-year clinico-epidemiological study of Histoid Hansen in India. *J Infect Dev Ctries* 2011;5:128-131.

Citing this article

Paudyal P, Shrestha S, Agarwal M, *et al.* Histoid leprosy on scrotal skin: an unusual case. *Int J Infect Microbiol* 2013;2(2):64-67.
