A rare case of genital porokeratosis associated with epididymo-orchitis

Sir,

Porokeratosis is a disorder of abnormal keratinization with varied clinical presentations in the form of localized and generalized disease.[1] Genital porokeratosis (GP) is a variant involving the scrotum, penis, buttocks, natal cleft, groins, and adjacent thighs. It is underreported as it mimics several venereal as well as nonvenereal dermatoses such as hypertrophic lichen planus, psoriasis, Bowen's disease, and dermatophytosis.[2] The exact etiology is yet unknown, but various trigger factors such as ultraviolet radiation, immunosuppression, drugs, malignancies, autoimmune diseases, and infections have been documented.[3] Here, we report a rare case of GP associated with bilateral acute on chronic epididymo-orchitis.

A 35-year-old immunocompetent male presented to the emergency department with fever and severe dragging scrotal pain for 3 days. No history of dysuria and burning or increased frequency of micturition was elicited. On further inquiry, he complained of dull aching pain in both testes and itchy lesions over the groin and scrotum for 1 year. Topical antifungal creams yielded no result. No other significant personal or family history was elicited.

On examination, multiple hyperpigmented papules and plaques with fine scaling and elevated peripheral rim, of size 1–3 cm in diameter, were visible discretely over the scrotum, bilateral inner thighs, and buttocks [Figure 1a and b]. Both testes were enlarged and tender, associated with thickened spermatic cords. Dermoscopy of plaques showed a well-defined lesion with a pale center, peripheral track-like border, and follicular plugging [Figure 2].

Investigation revealed normal hemogram, proteinuria (1+), and hematuria (1+). The urine culture was sterile. Ultrasonography showed tortuous and thickened bilateral spermatic cord with increased echogenicity and bilateral hydrocele. Skin biopsy from the border of the plaque depicted coronoid lamellae and pigmentary incontinence [Figure 3]. Based on these findings, a diagnosis of GP with chronic bilateral epididymo-orchitis with acute exacerbation was made. The patient was given topical 5-fluorouracil local application in the morning and topical tretinoin cream 0.025% cream at night locally along with oral isotretinoin 20mg at night. He was also advised tight scrotal support with oral levofloxacin 750mg once a day from the surgery unit. The patient is on regular follow-up and partial subsidence of lesions is seen after 1 month of treatment.

Chronic epididymo-orchitis is defined as scrotal pain lasting at least 3 months in duration and classified as inflammatory, obstructive, or epididymalgia.[4] Common infective causes include acute or preceding bacterial infections such as chlamydia and gonococcus in age <35 years, whereas coliforms and *Pseudomonas* are implicated in older males.[5] Our patient had features of chronic epididymitis, and acute exacerbation of pain with scrotal swelling suggesting acute epididymo-orchitis. The absence of urinary symptoms and sterile culture indicated that proteinuria and hematuria were secondary to epididymitis.

The infections found to be associated with porokeratosis are human immunodeficiency virus, herpes simplex, *Streptococcus*, hepatitis C, *Leishmania*, and human papillomavirus, but have been reported but in disseminated forms.[6] Chronic friction has been postulated to be a causative factor for GP, with no infectious agents demonstrated from the lesions. In a study, syphilis, condyloma acuminata, and folliculitis were associated in patients with GP.[7]

In our case, porokeratosis was present over the scrotum, buttocks, and adjacent thighs and was associated with acute on chronic bilateral epididymo-orchitis, which has not been reported in the literature. Acute epididymo-orchitis and porokeratosis are unrelated conditions, and GP was an incidental finding in our patient who presented with

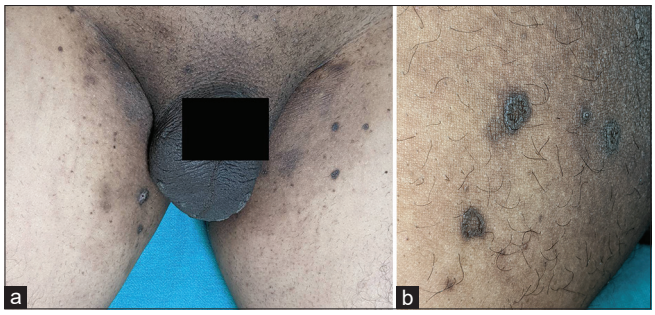

Figure 1: (a) Multiple keratotic papules and plaques with elevated scaly border and depressed center over the scrotum, and inner thighs bilaterally with scrotal swelling. (b) Well-defined keratotic plaques with raised border and depressed center

Figure 2: Hyperpigmented well-defined plaque with follicular plugging and double-marginated track-like border (DermLite DL4 ×10)

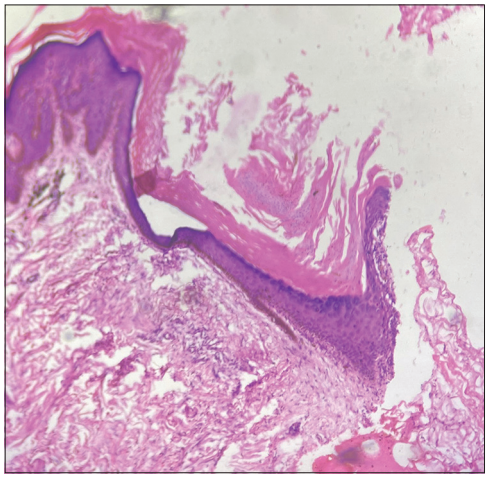

Figure 3: Coronoid lamella with pigmentary incontinence in the papillary dermis. (H and E ×400)

scrotal swelling and pain. Due to the simultaneous onset of lesions and chronic testicular pain, we suspect a common infectious etiology in both conditions, requiring further investigations such as polymerase chain reaction or nucleic acid amplification tests, which could not be done in our case due to logistic reasons. Thorough cutaneous and systemic examination and investigations in cases of GP should be done to establish if such association is definite or coincidental.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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