

Correspondence

Two Peas in a Pod: Exploring the Rare Co-existence of Genital and Extragenital Lichen Sclerosus Et Atrophicus in Two Patients

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Dear Editor,

Lichen sclerosus is a chronic inflammatory disease that primarily causes vulval and peri-anal lesion in prepubertal, peri- and post-menopausal women.^[1] Hallopeau described it as lichen planus et atrophicus in 1887. Now, the term has been changed to 'lichen sclerosus' since not all cases exhibit epithelial atrophy. The pathogenesis is unclear and female-to-male ratio ranges from 6:1 to 10:1.^[1] Lesions are mostly found in the anogenital region. Incidence of extragenital lichen sclerosus et atrophicus (LSA) is 15–20%,^[2] found mostly over the neck, shoulders, axillae, trunk and thighs. It is characterised by hyperpigmented/hypopigmented asymptomatic/itchy flat, polygonal papules and plaques with sclerosis and atrophy primarily affecting the vulva, perineum and perianal region in females and prepuce in males.

Patient 1: A 50-year-old male presented with progressively increasing asymptomatic atrophic depigmented macules and plaque with ill-defined borders over his right shin [Figure 1a] for the past 4 years. The patient also complained of thickening and depigmentation of the skin over his prepuce with mild itching and inability to retract the prepuce for the past 3 months [Figure 1b]. No erythematous papules over the glans or fissuring of the preputial skin present. There is no history suggestive of diabetes mellitus.

Patient 2: A 29-year-old male presented with progressively increasing asymptomatic, few ill-defined and few well-defined atrophic depigmented and few depigmented with hyperpigmentation in the centre macules over different parts of his body starting with flexor aspect of the upper limbs, then involving the axilla [Figure 1c], abdomen and trunk for the past 6 years. The patient also complained of thickening and depigmentation of skin with itching over his prepuce for the past 4 months with inability to retract the prepuce completely over his glans [Figure 1d]. There are no erythematous papules or fissuring of preputial skin present. There is no history suggestive of diabetes mellitus.

Skin biopsy from lesions over the shin [Figure 2a], prepuce [Figure 2b] and axillae [Figure 2c] revealed thin atrophied epidermis, vacuolar changes in stratum basale and follicular plugging. A cleft is evident in the dermo-epidermal junction due to dermal oedema and epidermal thinning. Superficial and deep perivascular infiltrate of lymphocytes and plasma cells with papillary dermal oedema and thickened dermal collagen is present, giving a hyalinised appearance. Dermoscopy of the extragenital lesion shows a structureless white to pinkish background,

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Figure 1: (a) Multiple atrophic depigmented macules and plaque with ill-defined borders over the right shin. (b) Depigmentation and atrophy of preputial skin. (c) Macules with central hyperpigmentation with atrophy and peripheral depigmentation with atrophy around the axilla. (d) Depigmentation and atrophy of preputial skin.

linear vessels and whitish comedo-like follicular plugging with perifollicular brown pigment [Figure 2d]. Based on the clinico-histopathological features, a diagnosis of genital LSA with extragenital LSA was made and treated with tacrolimus ointment 0.1% twice daily and clobetasol propionate 0.05% cream on lesions at night with mild improvement in lesions after 3 months.

LSA is a rare, chronic inflammatory dermatitis affecting both epidermis and dermis. Although aetiopathogenesis is unclear, genetic factors,^[1] autoimmune mechanism,^[2] borrelia infection,^[3] hepatitis C and androgen level irregularities are considered causative. LSA appears to have a correlation with autoimmune diseases with 25.5% of individuals having autoimmune disease and 42% with auto-antibodies in a study.^[3] Female-to-male ratio varies from 6:1 to 10:1.^[1] Prevalence is around 0.1% for children and 3% for women more than 80 years.^[1] Prevalence peaks between 8 and 14 years and 5–6th decades of life.^[3] Although it is mostly seen in the anogenital area (85–98%), it can also be seen in extra genital areas also (15–20%)^[2] mostly on the neck and upper arms.

Clinical manifestations include asymptomatic/itchy flat, opalescent papules that may cluster to form a plaque with

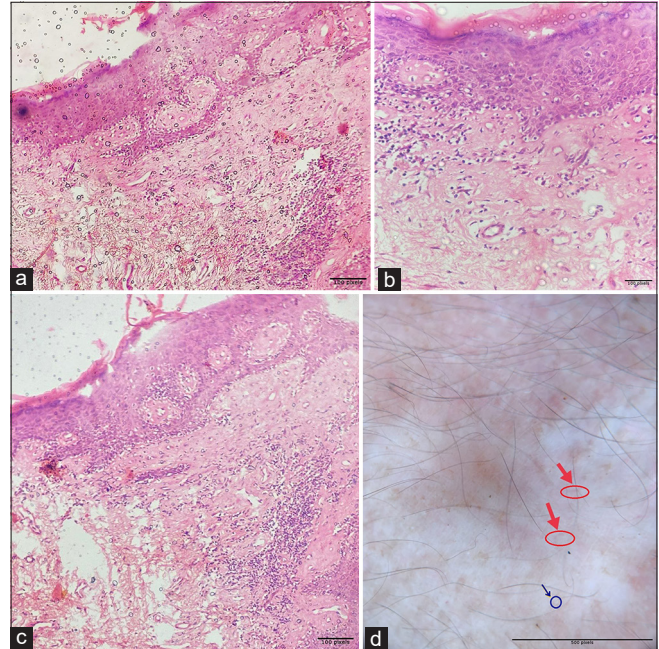


Figure 2: (a) Histopathology of lesions shows thin atrophied epidermis, vacuolar changes in stratum basale, dense superficial and deep perivascular infiltrate of lymphocytes and plasma cells in dermis with papillary dermal oedema and thickened papillary dermal collagen (H&E, ×400). (b) Histopathology of lesions shows thin epidermis, vacuolar changes in stratum basale, deep perivascular infiltrate of lymphocytes and plasma cells in dermis with papillary dermal oedema and thickened dermal collagen giving it a hyalinised appearance (H&E, ×400). (c) Histopathology of lesions shows thin epidermis, vacuolar changes in stratum basale with dermal oedema (H&E, ×400). (d) Dermoscopy of extragenital lichen sclerosus et atrophicus showing homogenous structureless white-to-pinkish background, linear irregular vessels (red arrows and circles) and whitish comedo-like follicular plugging with perifollicular brown pigment (blue arrow and circles) (Dermlite DL4, polarised mode, ×10).

sclerosis and atrophy involving the vulva, perineum and perianal region causing dyspareunia and dysuria in females (kraurosis vulvae). In males, it mainly involves the foreskin, causing sclerosis and narrowing, leading to phimosis (balanitis xerotica obliterans) and, later on, narrowing of the urinary stream and meatal stenosis if left untreated. Genital LSA is associated with the risk of malignant transformation. Extragenital LSA is usually asymptomatic, presents with whitish papules and plaques and is not associated with increased risk of malignant transformation to squamous cell carcinoma.^[4]

Treatment options include phototherapy (ultraviolet A1 [UVA1], psoralen and ultraviolet A [PUVA]), topical corticosteroids and topical tacrolimus. Therapies for generalised LS include PUVA/UVA1, hydroxychloroquine and methotrexate.^[3,5] Although uncommon, extragenital LSA should be considered in the clinical differential diagnosis of white patches over mucocutaneous regions.

We present this case due to the rarity of coexistence of genital LSA and extragenital LSA in a single patient with extragenital lesions being present over the shin in one patient which is an atypical location for extragenital LSA. Further, LSA is more common in females but both cases reported here are males. Early identification and treatment of genital and cutaneous lesions are crucial as they can cause considerable discomfort and morbidity, with a propensity for malignant transformation in genital lesions. Consequently, the case presented in this publication serves to enhance understanding of this uncommon condition.

Ethical approval: Institutional Review Board approval is not required.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for 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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