Blaschkoidal Lesion Over the Buttock in a Young Woman

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A woman in her 20s presented to the dermatology outpatient department with complaints of raised lesions in a linear distribution over the right buttock since 7 years of age. Lesions had progressed over the past few years to attain the current size. A depigmented remnant with hypertrophic scarring was seen in the lumbosacral region in the midline, which had resulted from an ablative attempt with carbon dioxide laser. There was no history of local trauma prior to the onset. The lesion was asymptomatic; history of bleeding or oozing was denied. Past history and family history were noncontributory. The lesion consisted of hyperpigmented punctate papules arranged in blaschkoidal distribution forming two streaks over the right buttock. Lichenoid hue was evident in the intervening skin [Figure 1a]. Dermoscopy revealed well-defined blue-gray papules with a peripheral keratotic rim, interspersed globules and blotches representative of dermal melanin, and a nonperipheral scale [Figure 1b]. Routine investigations did not reveal any abnormality. Histopathological examination was performed from one of the papules [Figure 2a and b].

What is the diagnosis?

Answer

Lichenoid porokeratosis.

Discussion

Differential diagnosis of blaschkoidal lichen planus, nevus comedonicus, linear porokeratosis, lichenoid porokeratosis, and porokeratotic adnexal ostial nevus were considered in this patient. Based on the clinical and histologic findings, a diagnosis of porokeratosis was established, and she was managed with topical 5% fluorouracil cream. Slight improvement in lesional thickness was observed at follow-up visit after 1 month. Porokeratosis presents as annular keratotic papules with a thready border that expands centrifugally. At least six clinical variants of porokeratosis are recognized. The thread-like raised hyperkeratotic border is characteristic, and its histologic correlate is cornoid lamella, a thin column of parakeratosis. The dermoscopic hallmark is a keratin rim; additional features include pigmentation along and within the rim, nonperipheral scales, and vascular structures.[1] Linear porokeratosis is a rare variant characterized by lesions of classical porokeratosis following the lines of Blaschko. Lesions arise frequently during infancy or childhood and most commonly involve extremities. Lichenoid porokeratosis is a subtype of linear porokeratosis characterized by prominent lichenoid tissue reaction on histopathology.[2] Prominent pigment incontinence observed on dermatoscopy and histopathology in the index case presents as a lichenoid hue on clinical examination that might resemble lichen planus.

Blaschkoidal lichen planus is characterized by linearly arranged violaceous, nonscaly, pruritic papules and plaques. The average age of onset was reported to be 33 years in a case series of 18 patients.[3] Lesions develop over weeks to months, and the course is self-limiting. Dermoscopic features include Wickham striae, vascular structures, and brown pigment dots. Although the lesions in the index case had a lichenoid hue on examination, asymptomatic nature, a non-remitting course, presence of surface scaling, lack of Wickham striae on dermoscopy, and coronoid lamella on histopathology negated the possibility of lichen planus.

How to cite this article: Mehta H, Thind A, Chatterjee D, Bishnoi A. Blaschkoidal lesion over the buttock in a young woman. Indian Dermatol Online J 2024;15:906-8.

Received: 20-Dec-2023. **Revised:** 24-Apr-2024. **Accepted:** 05-Jun-2024. **Published:** 08-Aug-2024.

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Access this article online

Website: https://journals.lww.com/idoj

DOI: 10.4103/idoj.idoj_953_23

Quick Response Code:

Nevus comedonicus is typically present at birth or develops in early childhood. Lesion appears as a collection of discrete, dilated follicular ostia filled with pigmented keratinaceous material. The site of involvement is the head and neck in most cases, and linear, segmental, and blaschkoidal patterns have been described.[4] Dermoscopy reveals brownish homogeneous structureless areas with multiple keratinaceous plugs. Honeycomb-like pitted scarring may occur as a sequalae. Histopathology is characterized by dilated follicular ostia devoid of hair shafts and filled with lamellated keratin. Punctate lesions in the index case lacked a central keratinaceous plug both clinically and on dermoscopy.

Porokeratotic adnexal ostial nevus (PAON) generally presents at birth and may have localized, generalized, segmental, or blaschkoidal distribution. Lesions are characterized by keratotic spines, punctate pits, and keratotic plugs. Histopathologically, parakeratotic columns arise from eccrine ducts and/or hair follicles. Later onset in childhood, absence of keratotic spines clinically, and presence of coronoid lamellae independent of hair follicles and eccrine ducts led to a diagnosis of porokeratosis rather than PAON.

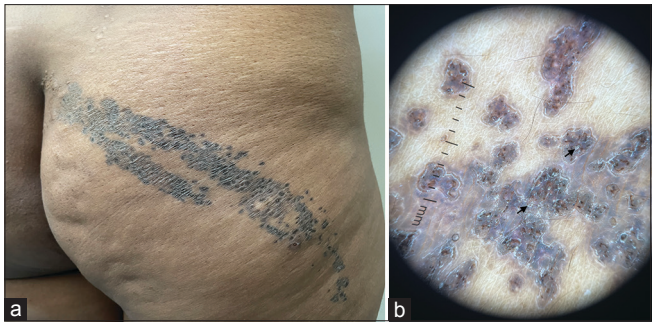

Figure 1: (a) Hyperpigmented punctate papules arranged in blaschkoidal distribution forming two streaks over the right buttock with a lichenoid hue in the intervening skin. (b) Well-defined blue-gray papules with a peripheral keratotic rim, interspersed globules and blotches representative of dermal melanin, and a non-peripheral scale on dermoscopy (black arrows) (Dermlite DL4, 10x)

Granular parakeratosis typically presents with pruritic papules or plaques in intertriginous areas and is characterized histopathologically by a thickened stratum corneum with abnormal retention of keratohyalin granules and parakeratosis.[5] However, in this patient, the lesions were chronic, asymptomatic, and located over the buttocks, which is not typical for granular parakeratosis. The absence of keratohyaline granules and the presence of a lichenoid infiltrate further argue against granular parakeratosis as the diagnosis.

The lichenoid infiltrate in this patient appeared less pronounced, likely attributed to the chronicity of the lesion. This case aligns with linear porokeratosis. Given the significant pigment incontinence and subtle vacuolar degeneration, it is reasonable to consider this presentation as lichenoid porokeratosis. Management options for porokeratosis include cryotherapy, topical 5-flurouracil, topical retinoids, topical imiquimod, and topical tacrolimus. Destructive procedures such as curettage, shave excision, dermabrasion, and linear excision have all been used with variable degrees of success. Resurfacing with carbon dioxide laser has been used successfully; however, one case report described recurrence of lesions over the entire treated area within 2 months.[6]

Development of squamous cell carcinoma has been well documented within lesions of porokeratosis, particularly in older patients, those of longstanding duration, and linear variants. Longitudinal follow-up is hence mandated in such patients.

Acknowledgement

We would like to thank the patient for providing consent for publishing the clinical images and case details.

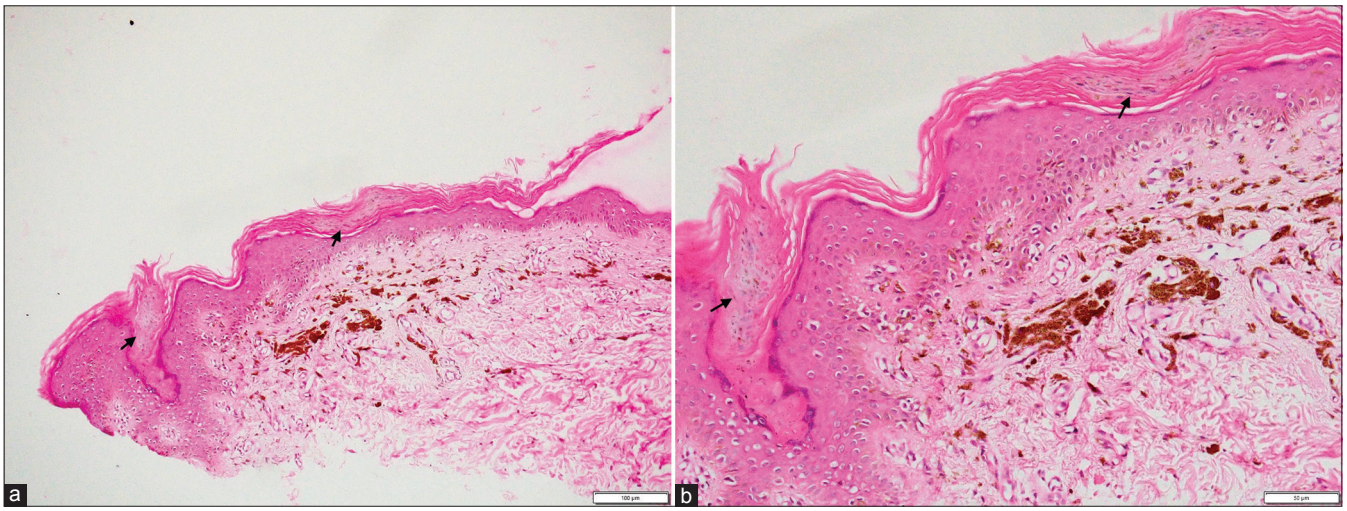

Figure 2: (a) Histopathological findings of focal epidermal parakeratosis (black arrows), hyperkeratosis, and basal cell vacuolization (H & E, 10×), (b) The epidermis shows distinct parakeratotic columns (black arrows). Melanin incontinence and moderate collection of perivascular inflammation composed of lymphocytes and plasma cells are evident in the dermis on higher magnification (H & E, 40×)

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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