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Keratosis lichenoides chronica: a case report with dermatoscopic and ultrasonographic analysis

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Abstract

We present the case of a 46-year-old woman with a long-standing history of linear erythematosquamous papules on the arms and thighs, which were unresponsive to treatment with topical corticosteroids. She also reported lesions typical of seborrheic dermatitis located on her face. After performing a clinical examination, dermatoscopy, high-frequency ultrasonography (HFUS), and a biopsy for histopathological examination, a diagnosis of keratosis lichenoides chronica (KLC) was obtained. Also known as Nekam disease, KLC is a rare, chronic inflammatory skin disease characterized by diffuse, linearly arranged erythro- or erythematosquamous papules with a thin, keratotic plug.

Key words: dermoscopy; keratosis lichenoides chronica; high-frequency ultrasonography.

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Introduction

Keratosis lichenoides chronica (KLC), also known as Nekam disease, is a rare chronic inflammatory skin disease characterized by diffuse, linearly arranged erythro- or erythematosquamous papules with a thin, keratotic plug.[1,2] Additionally, seborrheic-like lesions can occur on the face.[1] It has been widely discussed whether it is a separate entity or a subtype of lichen planus, lupus erythematosus, or lichen simplex chronicus.[2] Historically, it was named "*porokeratosis striata lichenoides*", although no cornoid lamella was found in the histopathological examination.[3,4] Herein, we present a case of KLC examined with dermatoscopy and high-frequency ultrasonography (HFUS). To date, we have not found any descriptions of the KLC ultrasound image in the literature.

Case Report

A 46-year-old woman presented with diffuse, linear erythematosquamous lesions located on the arms and thighs (Figure 1 a,b), which have been present for many years. She also reported recurrent episodes of seborrheic dermatitis on the face with a poor treatment outcome. Nail changes were not observed. Dermatoscopy (DermLite DL5, 10x magnification) revealed white scales covering purple structureless areas alongside dotted and linear vessels (Figure 1 c,d). HFUS examination (DermaScan C, Cortex, 20 MHz) demonstrated a hyperechogenic and irregular entrance echo with perpendicular acoustic shadowing, corresponding to the overlying scales of the lesions. Beneath the entrance echo, alternately arranged conical hypoechogenic structures were observed, with hyperechogenic linear projections extending into the dermis (Figure 2).

A punch biopsy was performed, and an acanthotic epidermis with verrucous hyperortokeratotic surface, irregular granular layer, and irregular rete ridges with hydropic degeneration of basal cells and occasional necrotic keratinocytes with dense band-like lymphohistiocytic infiltrate in the papillary dermis was seen in histopathology. Moreover, discrete parakeratosis within the hyperkeratotic plug in the dilated hair follicle, surrounded by inflammatory infiltrate, and inflammatory infiltrates surrounding dilated thin-walled blood vessels were also observed (Figure 3). The differential diagnoses were hypertrophic lichen planus and porokeratosis. The patient was initiated on systemic isotretinoin; however, treatment was discontinued after three months due to minimal clinical improvement and adverse effects, primarily severe skin dryness and irritation, which were poorly tolerated. She was subsequently started on UVA1 phototherapy but was only able to complete 10 sessions owing to personal circumstances. At follow-up, the patient expressed acceptance of her condition and declined further therapeutic interventions.

Discussion and Conclusions

KLC is an ambiguous dermatologic condition, with approximately 80 cases described in the literature.[1] The treatment modalities used for this disease include retinoids or phototherapy. However, evidence regarding their efficacy remains limited, pri-

marily derived from individual case reports, with no randomized controlled trials available.[3] Furthermore, there are only a few reports on noninvasive skin imaging in KLC; in two of them, dermoscopic images were reported, and in one of them, the findings in reflectance confocal microscopy were described. On dermoscopy, a desquamative plaque with irregular edges, arranged in a hyperpigmented reticular pattern with superficial white scales on an erythematous pink base, was seen in one of the reports.[5] The second case revealed thick whitish lines, dotted and linear vessels, and brown-gray irregular and perifollicular granules and dots.[6] In our case, in HFUS, we observed a hyperechogenic entrance echo related to the verrucous hyperortokeratotic surface of the lesions and hypoechogenic, conical structures related, among others, to the changes in the deeper part of the epidermis, as well as dense band-like lymphohistiocytic infiltrate in the papillary dermis. The linear, ingrowing hyperechoic lines may represent a distinctive ultrasonographic feature of KLC, potentially corresponding to dilated hair follicles filled with hyperkeratotic and parakeratotic material. This

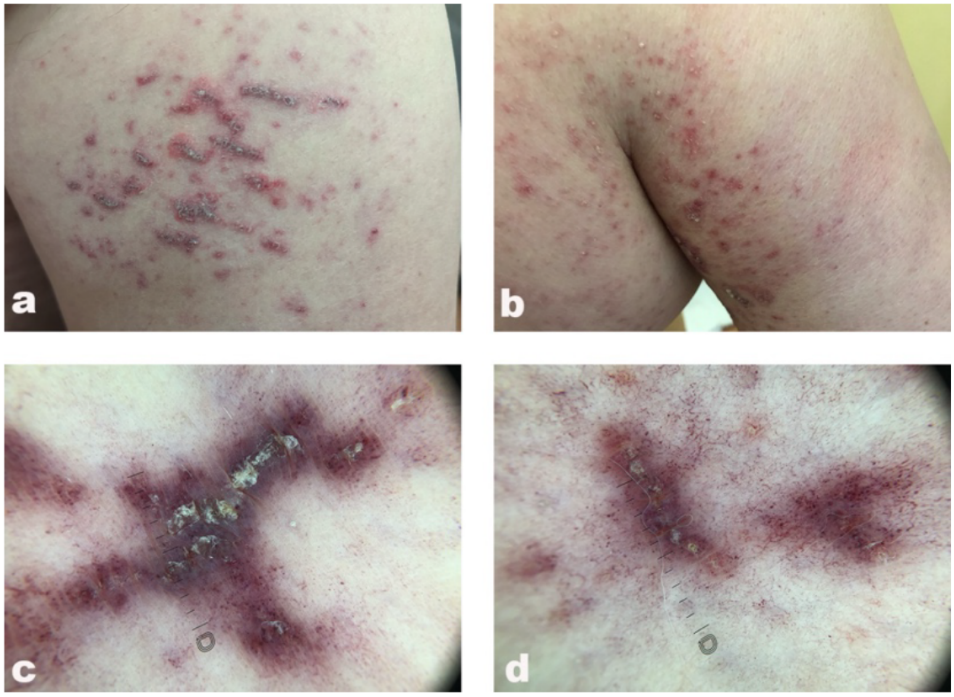

Figure 1. Erythrosquamous lesions clinically seen on **a**) the thigh and **b**) the upper extremity. **c, d)** Dermatoscopy revealed white scales arranged linearly, covering purple structureless areas alongside dotted and linear vessels.

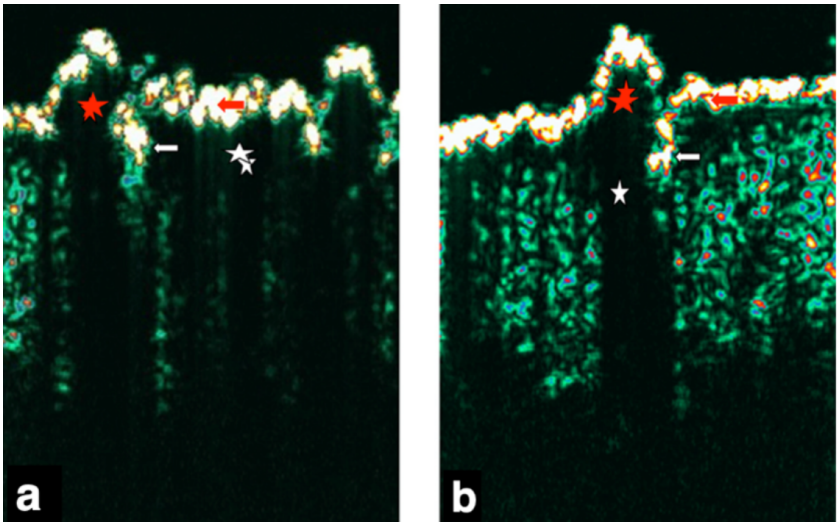

Figure 2. 20 MHz scans of KLC with hyperechogenic and irregular entrance echo (red arrow), perpendicular shadowing corresponding to the scales covering the lesions (white star), and the alternately arranged conical hypoechogenic structures (red star) and hyperechogenic lines ingrowing to the dermis (white arrow).

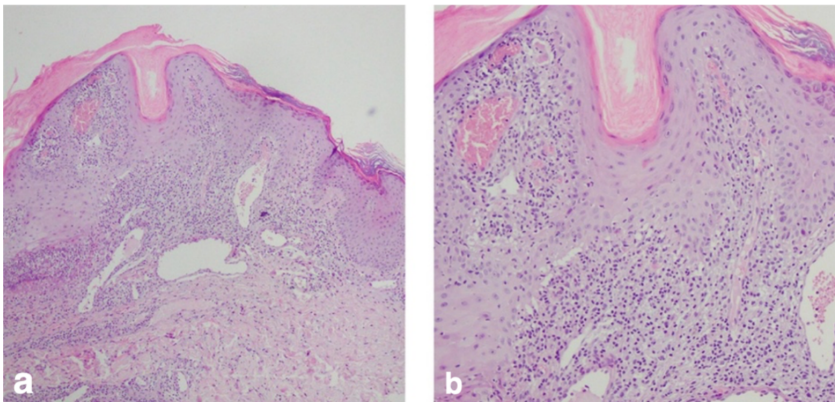

Figure 3. Histopathology. **a**) Acanthotic epidermis, irregular rete ridges, hyperkeratotic surface with follicular plug, dense band-like lichenoid infiltrate, and dilated blood vessels surrounded by inflammatory infiltrates (original objective magnification 4x, H&E); **b**) hydropic degeneration of basal keratinocytes, cytoid body, parakeratosis in the hair follicle, and lympho-histiocytic infiltrate (original objective magnification 10x, H&E).

finding could aid in differentiating KLC from hypertrophic lichen planus; however, validation in a larger patient cohort is necessary.

The presence of scales covering the lesional skin is responsible for perpendicular shadowing and impairs the ability to assess the boundaries of the lesion. While histopathology remains the gold standard in the diagnosis of KLC, it is not always evident. In most cases, this lichenoid eruption resembles verrucous lichen planus, with some exceptions like occasional parakeratosis, perifollicular/acrosyringotropic, or perivascular inflammatory infiltrates.[7] The currently available therapies for KLC include systemic retinoids (isotretinoin and acitretin) and phototherapy, which have been described to have a satisfying efficacy in some patients.[1] Furthermore, resistance to dapsone, systemic steroids, methotrexate, and cyclosporine has also been reported.[1,8] Recently, Tang *et al.* reported a case in which a Janus kinase inhibitor (upadacitinib 15 mg/day) was used in a KLC patient. A near-complete clearance of the lesions on his limbs was observed after 5 months of therapy.[8] Additional non-invasive techniques, such as HFUS and dermatoscopy, can assist physicians in making a preliminary diagnosis. Although diagnosing this condition remains challenging, identifying its specific dermatoscopic and HFUS features would require a larger patient cohort – something difficult to achieve given the rarity of this dermatosis.

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Consent for publication: the patient gave her written consent to use her personal data for the publication of this case report and any accompanying images.

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