Our Dermatology Online

Case Report

Recurrent unilateral linear capillaritis: A very rare variant of pigmented purpuric dermatosis

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ABSTRACT

Pigmented purpuric dermatosis is a disease characterized with localized purpuric lesions. Six different clinical types have been described. There are also unclassified variants including rare granulomatous and unilateral linear capillaritis. Unilateral linear capillaritis is characterized by a very rare, unilateral linear or segmental purpuric macula. A 18-year-old female patient was presented with a linear purpuric and petechial patch and macules on the left arm. Unilateral linear capillaritis is characterized with benign, recurrent purpuric lesions in children and young adolescants that may be presented with spontaneous regression.

Key words: Linear; Pigmented; Unilateral

INTRODUCTION

Pigmented purpuric dermatosis (PPD) is usually characterized with localized purpuric lesions in the lower extremities, and it consists a group of dermatosis with similar histopathological findings caused by an unknown etiology [1-3]. Six different clinical types have been described: Progressive pigmented purpuric dermatosis (Schamberg's purpura), Purpura annularis telangiectoides (Majocchi purpura), lichen aureus, pigmented purpuric lichenoid dermatosis of Gougerot and Blum, itching purpura and eczematia-like purpura of Doucas and Kapetanakis. There are also unclassified variants including rare granulomatous and unilateral linear capillaritis [2]. Unilateral linear capillaritis (ULC), also known as segmental pigmented purpura or Quadrantik capillaropathy, is characterized by a very rare, unilateral linear or segmental purpuric macula. It is a benign condition presented with spontaneous regression [2,4-6]. As far as we know, 16 cases were reported so far [2]. Since its incidence is rare, our 18-year-old female patient is presented here.

CASE REPORT

A 18-year-old female patient was admitted to our outpatient clinic with complaints of redness on the left arm in November 2017. She had complaints for 3 years and it repeats every 2 months. The lesions were healing in 5-7 days. There were no pain, itching and burning. No family history. She has mitral valve prolapse and she has taken beta blockers (metoprolol) since 2012.

On dermatological examination, she had a linear pigmented rash on the left arm. The eruption consisted of linear purpuric and petechial patch and macules and did not blanch with pressure from a diascope (Figs. 1 and 2). Hemogram and biochemical tests were normal. The clinical diagnosis was felt to be unilateral linear capillaritis. The patient went on to have a 4mm punch biopsy from the left arm.

Histopathological examination revealed perivascular lymphocytic infiltrate and erythrocyte extravasation in the upper dermis (Fig. 3). Based on clinical and histological findings, the patient was diagnosed

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Figure 1: Linear pigmented purpuric rash on the left arm.

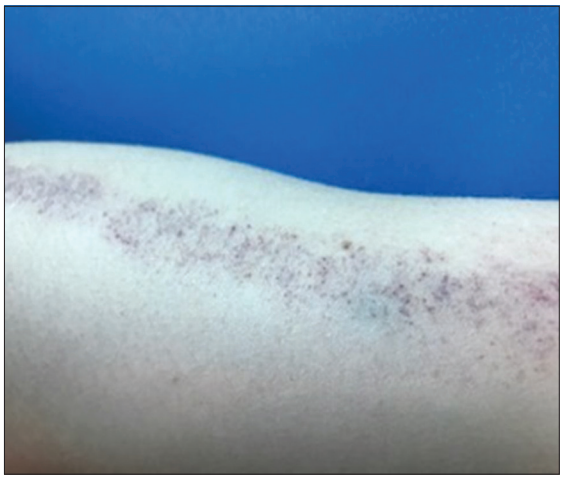

Figure 2: A closer view of the lesion.

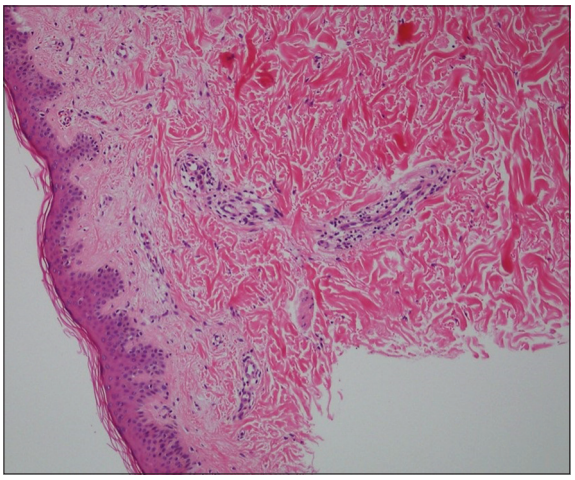

Figure 3: Under the epidermis, perivascular lymphocytes and extravascular erythrocytes.

as unilateral linear capillaritis. Treatment was not recommended because the symptoms spontaneously recovered in about 5 days. The patient was followed up.

DISCUSSION

Higgins et al. have identified a female patient with so-called quandrantic capillaropathy [5] in 1990, for the first time. Riordan reported 4 patients with so-called unilateral linear capillaritis in 1992 [7]. PPD is presented usually in middle and older ages, involving lower extremities symmetrical. ULC patients are younger and the lesions disappear spontaneously in about 3 years. The cause of the unilateral and linear involvement is unknown [8].

The suspected etiology include venous hypertension, exercise, capillary fragility, focal infections, alcohol drugs are implicated [9]. Aspirin, carbomal, thiamin, meprobamate, carbamazepine, reserpine, acetaminophen, glipizide, hydralazine are the suspected drugs [8]. The drugs are reported to be the trigger, especially for schamberg disease [1]. Only one of the reported ULC case had been associated with Aspirin [8]. Our patient uses beta-blockers but since she had spontaneous resolution, her condition was not found to be associated with the drugs histopathologically.

So far, 9 (56.25%) of the patients were male and 7 (43.75%) were female. Its incidence is more common in men, but the difference is not significant. The age of the patients range 5 - 48 years. Half of the patients are children. There are two peak ages: 5-15 years (mean 10.12 years) and 23-56 years (mean 35 years). It is usually asymptomatic, Mar reported mild pruritus in two patients [4]. Lower extremity involvement was observed in 11 (68.7%), upper extremity involvement was observed in 4 (25%) and body involvement was observed in 3 (18.7%) patients (1 patient had only body involvement and 2 patients had body involvement with lower extremity involvement). The lesions are usually in segmental configuration, with less linear involvement [2,10]. Since spontaneous regression is observed in a few months or years, generally treatment is not required. Topical steroids, hydroxychloroquine or PUVA may be given if required [8].

The differential diagnoses should include angioma serpiginosum, unilateral naevoid telangiectasia, and lichen aureus firstly [4,11]. Lichen aureus is differentiated by having no band-style lichenoid infiltration on dermis [11].

In conclusion, ULC is a pigmented purpuric dermatosis characterized with benign, recurrent purpuric lesions in children and young adolescants that may be presented

with spontaneous regression mostly involving lower extremities with segmental or linear lesions.

Consent

The examination of the patient was conducted according to the Declaration of Helsinki principles.

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