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Case Report

A Rare Case of Erythema Elevatum Diutinum with Blister

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Abstract

We report a rare case of erythema elevatum diutinum with blisters, a 58-year-old woman presented with recurrent erythema, nodules, plaques, and blisters on her face, trunk, and extremities without no cause. The biopsy of the blisters on the left lower leg showed Leukoblastic vasculitis and the biopsy of the plaque on the dorsal right hand showed persistent elevated erythema tissue. The vesiculobullous appearance of EED is extremely rare, and the histopathological in both the acute and chronic stage found on skin biopsy are more worth reporting.

Keywords: Erythema; Nodules; Blisters; Neutrophils

Introduction

Erythema elevatum diutinum is a rare localized variant of cutaneous leukoblastic vasculitis, characterized by violet, reddish-brown papules, plaques or nodules, occasionally bullae and ulcers, rare blisters or bullous variants. We report a rare type of erythema elevatum diutinum with vesiculobullous.

Case Report

A 58-year-old woman presented with a two year history of papules, nodules and blisters on the trunk and limbs, especially on the extensor side of limbs and joints. Accompanied by mild pruritus. The patient was in good general health, with no history of chronic diseases. The lesions were partly subsided after treatment with Tripterygium wilfordii. Five months ago, the lesion reappeared again.

Physical examination reveals multiple erythema, papules in the face, waist, limbs, with dense blisters on her lower legs, the violet patches, nodules, were distributed on the extensor sides of both hands and feet. Lower lip mucosa can be seen soy-sized ulcer, and there was an ulcer on the mucosa of the lower lip (Figure 1). Two skin biopsies were performed. The pathological findings from the blister on the left Lower leg showed hyperkeratosis of the epidermis, edema of cells in the spinous layer, fibrinoid necrosis in the middle and upper dermis, infiltration of neutrophils and lymphocytes, and a large number of nuclear dust and cells were observed. The above changes were mainly in the superficial dermis. (Figure 2A). The nodule on right hand biopsy showed epidermal hyperkeratosis accompanied by parakeratosis, hyperplasia and hypertrophy of the spinous layer, elongation and broadening of the epidermal process, scattered or large patches of neutrophils, lymphocytes and histiocytes infiltrate around the blood vessels in the whole dermis, and scattered eosinophils are present (Figure 2B). Direct immunofluorescence and indirect immunofluorescence were both negative. Abnormal laboratory examination showed indicated elevatied eosinophils number of 1.381×0^9/L↑ (reference range, 00.5×10^9/L), ANA:1:1000, karyotype centromeric type, anti CENPB +, AMA-M2 positive; cryoglobulin positive, anti-M2-3E positive, gp210 positive, Bone marrow showed eosinophils increased. Laboratory investigation including liver and kidney function, Rheumatoid factor, serum protein electrophoresis, ASO, urine routine, C-reactive protein, tumor markers, C3 and C4 were all within normal limits. HIV, HBV, HCV, breast molybdenum target, chest CT, ECG were either negative or normal. The diagnosis of Erythema Elevatum Diutinum (EED) is made by skin manifestations and histopathological examination, patients was received the injection of 1mg compound betamethasone , in view of the nodules in giving depot was detected in the lesions of 1mg intramuscular injection treatmen, clinical symptoms were improved in the next 1 weeks, The eosinophils were reduced to normal. Abnormal autoantibody spectrum of the patient was considered to be primary biliary cholangitis, and liver puncture examination was suggested for further clarification, but the patient refused temporarily. The patient's skin lesions were not recurred during the current follow up.

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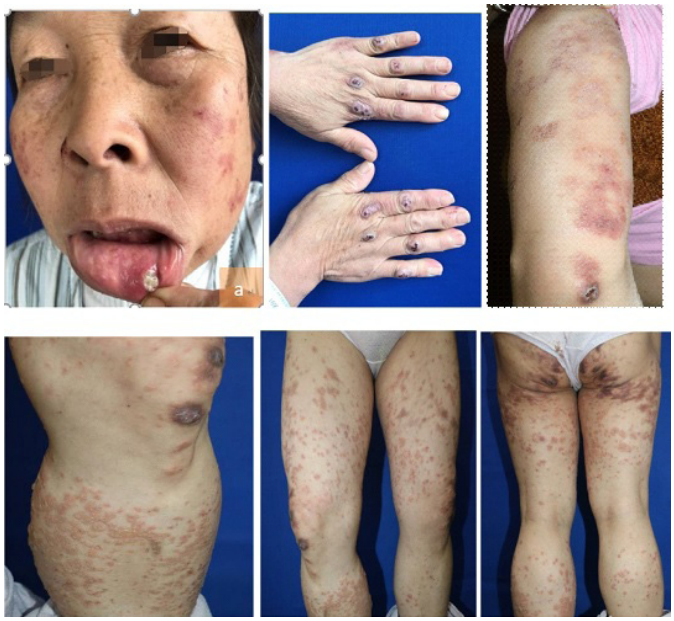

Figure 1: Infiltrative rash on the legs at presentation.

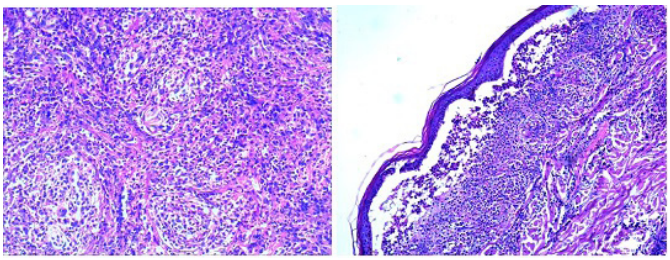

Figure 2: A) Hperkeratosis of the epidermis, edema of cells in the spinous layer, fibrinoid necrosis in the middle and upper dermis, Inflammatory cells infiltrate the superficial dermis (H&E, original magnification x 40). **B)** Infiltration of neutrophils and lymphocytes, and a large number of nuclear dust and cells were observed (H&E, original magnification x 100).

EED is a rare localized variant of cutaneous leukocytic vasculitis, which usually occurs between 30 and 60 years with no vary by sex. The typical lesions characterized by violet, red-brown papules, plaques or nodules. The lesions are mostly present on the extensor surface of limb and joints. and the trunk is often not involved. However, some atypical sites such as the penis [1] oral mucosa [2,3] cornea [4] and larynx [5] have been reported. Early manifestations may be ecchymosis, with occasional bullae, ulceration, and rare blisters or bullae variants. Histologically [6], a spectrum from leukocytoclastic vasculitis to vessel occlusion and dermal fibrosis was seen. EED disease may be idiopathic, but it may also be an underlying systemic disease, including systemic infection (hematological disorders and Autoimmune and inflammatory diseases [7] and certain malignancies (B-cell lymphoma, squamous cell carcinoma, breast carcinoma) [8]. It is important to conduct a thorough examination of the whole body after the discovery of EED patients to exclude the potential disease. The laboratory examination of the patient found ANA positive, AMA-M2 positive, anti-M2-3E antibody positive, GP210 positive, the above indicators more occurred in patients with primary bile cholangitis, primary bile cholangitis pathogenesis is related to autoimmunity, Therefore, persistent erythema bulge in patients may be related to autoimmune diseases, and no case related to EED and primary biliary cholangitis has been reported. The patient was cryoglobulin positive, may also be related to EED.

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