

## Bullous eosinophilic cellulitis with subcorneal pustules

Sir,

Eosinophilic cellulitis, or Wells syndrome, is an uncommon inflammatory dermatosis of unclear pathogenesis, often characterized by erythematous, edematous plaques that resemble acute bacterial cellulitis or urticaria. Recently, a variety of clinical lesions have been reported in patients with eosinophilic cellulitis, including papules, nodules, vesicles, and, rarely, bullae. However, only a few cases of non-bullous eosinophilic cellulitis with subcorneal pustules have been reported.<sup>[1,2]</sup> Here, we describe a patient with bullous eosinophilic cellulitis showing subcorneal neutrophilic and eosinophilic pustules.

A 39-year-old Japanese female presented with a 1-month history of severely pruritic, erythematous vesicular eruptions on her extremities and a 1-week history of erythematous papular lesions on her trunk, which did not respond to oral antihistamines and a topical steroid. There was no history of any antecedent drug intake, chronic sinusitis or asthma.

On physical examination, there were well-demarcated, oval, edematous, erythematous plaques, vesicles, bullae and pustules on the dorsa of the hands, forearms, and soles [Figure 1a]. The patient had tenderness in these areas. Moreover, multiple erythematous lesions with papules, pustules and crusts were present on the trunk and extremities [Figure 1b]. She did not have any systemic symptoms such as malaise, fever or arthralgia. Laboratory studies were significant for a white blood cell count of  $8400/\mu\text{L}$  with 15% eosinophils. All other laboratory data, including autoantibodies, were within normal limits. Bacterial culture from a pustule was sterile. A diagnosis of eosinophilic cellulitis was suspected; however, other clinical differentials considered were pyoderma

gangrenosum, Sweet's syndrome, and contact dermatitis with id reaction.

Two skin biopsies were performed, one from the bullous lesion on right forearm and another from the erythematous plaque on the abdomen [Figure 1a and b]. Histological examination of the forearm lesion showed subcorneal pustules containing mainly neutrophils, severe dermal edema resulting in subepidermal blisters and dense infiltration consisting of equal numbers of eosinophils and neutrophils throughout the dermis [Figure 2a and b]. Flame figures were observed in the dermis[Figure 2c]. Biopsy from the abdominal lesion showed subcorneal pustules containing predominantly eosinophils [Figure 2d]. Perivascular and interstitial infiltrates of eosinophils and lymphocytes as well as flame figures were found in the dermis [Figure 2e]. There was no vasculitis.

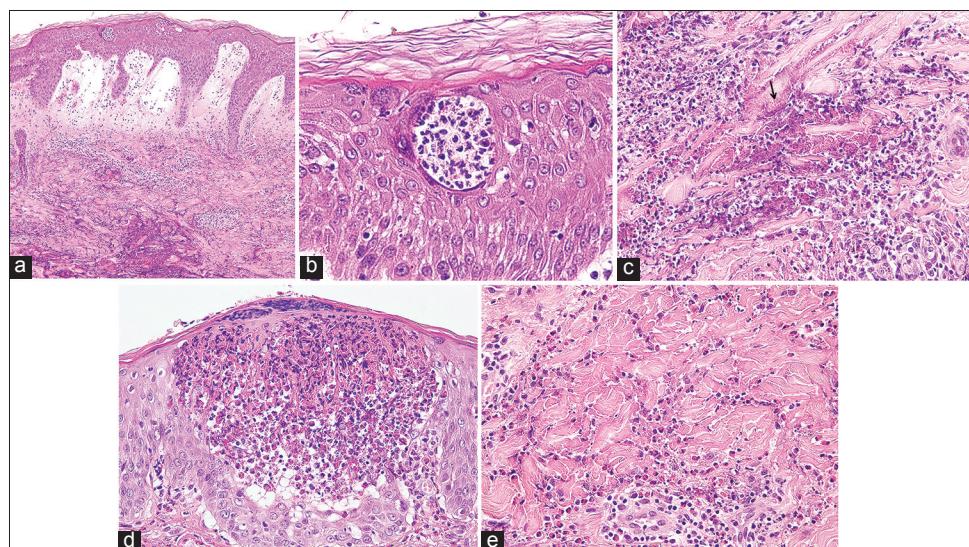


**Figure 1:** (a) Edematous erythematous plaques, vesicles, bullae, and pustules on the dorsum of the hand and distal forearm. (b) Erythematous plaques with papules, pustules, and crusts on the abdomen

Direct immunofluorescence was negative for immunoglobulin G (IgG), IgA, IgM, and C3. Indirect immunofluorescence using monkey esophagus and salt-split human skin as substrates showed no circulating IgG or IgA autoantibodies.

Based on these findings, a diagnosis of bullous eosinophilic cellulitis was made. The patient was commenced on prednisolone 0.5 mg/kg daily and fexofenadine 60 mg twice daily combined with a topical steroid. Clinical improvement of the skin lesions was observed 2 weeks after treatment. Prednisolone use was tapered and discontinued after 9 weeks. During a six month follow up period, there was no clinical relapse or eosinophilia.

Subcorneal pustules are very rare in eosinophilic cellulitis, and have previously been described only in two cases of eosinophilic cellulitis;<sup>[1,2]</sup> however, both these cases were of non-bullous eosinophilic cellulitis. We were unable to find any previous report of bullous eosinophilic cellulitis, which histologically showed subcorneal pustules. Takata *et al.*,<sup>[1]</sup> reported cutaneous botryomycosis followed by non-bullous eosinophilic cellulitis with sterile subcorneal pustules. Chao *et al.*,<sup>[2]</sup> reported non-bullous eosinophilic cellulitis with subcorneal pustules, diagnosed as an id reaction, after a molten aluminium burn. Although two cases of bullous eosinophilic cellulitis with pustules have been reported,<sup>[3,4]</sup> histological features of the pustules were not described.



**Figure 2:** (a) The bullous lesion shows subcorneal pustules, subepidermal blisters, papillary dermal edema, and dense infiltration of neutrophils and eosinophils involving the full thickness of the dermis (H and E,  $\times 100$ ). (b) Subcorneal pustules containing mainly neutrophils (H and E,  $\times 400$ ). (c) Flame figures in the dermis (H and E,  $\times 400$ ). (d) The erythematous plaque shows subcorneal pustules containing many eosinophils (H and E,  $\times 400$ ). (e) Aggregates of eosinophilic granules and nuclear fragments around collagen bundles in the dermis. Perivascular and interstitial infiltrate of eosinophils and lymphocytes as well as flame figures in the dermis (H and E,  $\times 400$ )

The important finding in this case was that subcorneal neutrophilic pustules and subcorneal eosinophilic pustules were observed simultaneously; however, it is difficult to explain the findings. We think that these two forms of pustules might be due to different pathogenic mechanisms or simply due to the time course of each lesion. A biopsy from the abdominal lesion performed 1 week after the appearance of the erythematous papules showed subcorneal eosinophilic pustules. By contrast, a biopsy from the right forearm performed 1 month after the occurrence of the vesicular eruptions displayed subcorneal neutrophilic pustules. Indeed, Takata *et al.*<sup>[1]</sup> performed a biopsy 3 weeks after onset and diagnosed eosinophilic cellulitis with subcorneal eosinophilic pustules, whereas Chao *et al.*<sup>[2]</sup> performed a biopsy after 6 weeks and diagnosed eosinophilic cellulitis with subcorneal pustules containing both neutrophils and eosinophils. Eosinophils can release interleukin (IL)-8, which exhibits chemotactic activity for neutrophils.<sup>[5]</sup> We postulate that subcorneal eosinophilic pustules could be sequentially replaced by subcorneal neutrophilic pustules.

**Taisuke Kamiyama, Hideaki Watanabe,  
Hirohiko Sueki**

Department of Dermatology, Showa University School of Medicine,  
Tokyo, Japan

**Address for correspondence:** Dr. Hideaki Watanabe, Department  
of Dermatology, Showa University School of Medicine, 1-5-8,  
Hatanodai, Shinagawa-ku, Tokyo 142-8555, Japan.  
E-mail: hwatanabe@med.showa-u.ac.jp

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