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CASE REPORT

Acute Localized Exanthematous Pustulosis on the Face

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Acute localized exanthematous pustulosis (ALEP) is a localized variant of acute generalized exanthematous pustulosis, which is characterized by the eruption of multiple scattered pustules following drug administration. A 26-year-old woman presented with multiple erythematous pustules on her face, which had appeared three days after taking antibiotics. Histopathological findings showed subcorneal pustules and mixed inflammatory cell infiltration in the dermis. The pustules were resolved within 2 weeks after the patient discontinued the antibiotics. Herein, we present a case of a woman with a cutaneous drug reaction consistent with ALEP that occurred subsequent to administration of antibiotics. **(Ann Dermatol 23(S3) S368～S370, 2011)**

-Keywords-
Acute generalized exanthematous pustulosis, Acute localized exanthematous pustulosis, Antibiotics

INTRODUCTION

Acute generalized exanthematous pustulosis (AGEP) is a severe drug reaction that is characterized by the eruption of dozens to hundreds of scattered pustules following drug administration[1]. A localized variant of this pustular reaction called "acute localized exanthematous pustulosis (ALEP)" rarely occurs.

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Herein, we present a case of a woman with a cutaneous drug reaction consistent with ALEP that occurred subsequent to administration of antibiotics.

CASE REPORT

A 26-year-old woman presented with an acute outbreak of asymptomatic multiple pustules and surrounding erythema affecting the face. One week ago, she took antibiotics (cephalosporine, bactrim, and vancomycin) for the treatment of otitis media. After three days, she reported subjective fever, fatigue and a chilling sensation and pustules with surrounding erythema were observed on her face. The pustules were superficial and nonfollicular with a surrounding erythema. Her white cell count was 1.77×10^9 L^{-1}, with an increased neutrophil count (80.4%). The erythrocyte sedimentation rate (ESR) and c-reactive protein (CRP) were elevated (ESR: 55 mm, CRP: 43.91 mg/dl). The blood chemistry, liver function, and renal function tests were within the normal limit. Cultures were negative for bacteria. Histopathological findings showed subcorneal pustules with scattered apoptotic keratinocytes adjacent to the pustules. The papillary dermis was edematous and prominent mixed inflammatory cells infiltrated in the dermis, with leukocytoclastic vasculitis. She was discontinued use of the antibiotics and was treated with systemic corticosteroid (methylprednisolone 8 mg/day). The pustules were rapidly resolved over several days. Two weeks later, most lesions disappeared and small hemorrhagic crusts and pigmentation remained. The scheduled patch test to identify the causative drug was canceled due to the patient's personal reason.

DISCUSSION

AGEP is an uncommon but well-known cutaneous drug reaction that is characterized by the eruption of dozens to hundreds of scattered pustules following drug admi-

nistration. The pustules are acute, generalized, nonfollicular and often coalesce on an edematous, erythematous background. The eruption can be accompanied by fever and neutrophilic leukocytosis[1]. Antibiotics are the most commonly drugs that causes these symptoms although non-steroidal anti-inflammatory drugs, antifungals, calcium antagonists, and antiulcer drugs may also trigger this response[2]. The pathophysiological mechanism of these symptoms remains largely unknown. But previous studies in patients with AGEP have revealed a high rate of positive patch test responses and in vitro lymphocyte proliferative responses compared with other drug eruption. This strongly suggests that this adverse cutaneous reaction occurs by a drug-specific T-cell-mediated process[3]. These specific T-cells produce large amounts of cytokines, which contribute to the accumulation of neutrophils[4].

ALEP is a less common form of pustular drug eruption, in which lesions are consistent with the characteristics of AGEP but are typically localized to the face, neck or chest. Prange[5] and colleagues first defined ALEP in 2005. They described a woman whose symptoms fit the criteria of AGEP but whose lesions were localized to the face. They suggested that ALEP was a variant of AGEP both clinically and histopathologically. To the best of our knowledge, only nine separate case of this variant has been reported. Two cases occurred following administration of amoxicillin-clavulanic acid[5,6], four following administration of amoxicillin only[6-9], and one after administration of levofloxacin[10]. Other causative drugs aside from antibiotics include ibuprofen[3] and paracentamol in pregnant woman[11]. In reported cases, the lesions are consistent with the characteristics of AGEP but are localized typically to the face, neck, and chest.

In the present case, the Sweet syndrome (acute febrile neutrophilic dermatosis) should be differentiated. Typical findings of the Sweet syndrome include the sudden appearance of a fever, increased neutrophilic count, erythematous papules, pustules on the head, neck, and extremities and infiltration of neutrophils throughout the dermis. However, in the Sweet syndrome, the lesions are usually tender and there is no evidence of leukocytoclastic vasculitis in the dermis[12]. Histologically, the subcorneal pustules of ALEP resembled those of pustular psoriasis or subcorneal pustulosis. Recent drug administration history, no personal history of psoriasis, more acute course and rapid spontaneous healing allowed us to differentiate the diagnosis from other pustular dermatoses[13].

The most important aspect of treatment is the immediate withdrawal of the suspected agent and supportive therapy. Other specific treatments are not usually needed, because ALEP is a self-limited disease with a favorable prognosis in most cases[3]. However, topical or systemic corticosteroids can be helpful because of their anti-inflammatory properties[14].

In our case, the lesions developed rapidly after administration of antibiotics and were resolved within several days of discontinuing the treatment. Based on this histologic findin, the symptoms appeared to correspond to AGEP, which results in unusual characteristics that are limited on the face. All antibiotics that had been used in this case can lead AGEP. However, only two cases attributed to vancomycin have been reported[15]. Cephalosporins are relatively common causative drugs for AGEP, but interestingly not sulfonamides, who have a high potential of causing other drug eruptions[1]. Therefore, we assumed that cephalosporins were the causative drug in this case.

Herein, we present a case of a woman with a cutaneous drug reaction consistent with ALEP that occurred subsequent to administration of antibiotics. Further study may be required to facilitate the correct diagnosis and treatment for this rare disorder.

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