A Case of Neutrophilic Eccrine Hidradenitis in an Infant

Kyoung Jin Kim, M.D., Jeong Yeob Lee, M.D., Jee Ho Choi, M.D., Kyung Jeh Sung, M.D., Kee Chan Moon, M.D., Jai Kyoung Koh, M.D.

Department of Dermatology, Asan Medical Center, College of Medicine, University of Ulsan, Seoul, Korea

Neutrophilic eccrine hidradenitis (NEH) is a rare neutrophilic dermatosis which is characterized by the erythematous papules or plaques with a neutrophilic infiltrates around eccrine glands and coils. In 90% of cases, NEH develops in patients who have received chemotherapy for malignant diseases, but other drugs, infections, and paraneoplastic phenomena are considered as possible etiologic factors. Furthermore, NEH is known to be extremely rare in infants. Herein we report a case of NEH in a nine-month-old infant with acute myelogenous leukemia (AML), which lesions appeared on the extremities including palms and soles before starting chemotherapy. (Ann Dermatol 14(2) 106-109, 2002).

Key Words : Neutrophilic eccrine hidradenitis (NEH), Infant, Before chemotherapy

CASE REPORT

A nine-month-old female infant presented with multiple, erythematous, tender maculopapules and patches on the upper and lower extremities for two months. Two months earlier, cough and easy bruisability developed and several erythematous infiltrative papules appeared on both dorsum of hands and feet one week later. Thereafter fever, decreased appetite, and dyspnea presented. She was diagnosed as a pneumonia with sepsis and treated with antibiotics, but there was no clinical improvement. The skin lesions were more erythematous and wide spread to both forearms, legs, palms and soles. Laboratory investigations showed the following results or findings: leukocytes, $31.2 \times 10^3 / \text{mm}^3$ (normal, $4.0 \text{ to } 10.0 \times 10^3 / \text{mm}^3$), with differential count of monocytes, 20%, lymphocytes, 21%, atypical lymphocytes, 1%, segmental neutrophils, 49%, hemoglobin, 10.9 g/dl (normal, 12 to 16 g/dl), platelet, $170 \times 10^3 / \text{mm}^3$ (normal, 150 to $350 \times 10^3 / \text{mm}^3$), CRP, 19.11mg/dl (normal, 0-0.6), reticulocytes, 46.5% (normal, 0 to 17%), and peripheral blood smear showed immature and large dysplastic monocytes, suggestive of myelodysplastic syndrome. A bone marrow biopsy was done and she was diagnosed as AML, M4.

The erythematous skin lesions increased in number
and the patient was referred to the dermatologic clinic. Skin examination revealed multiple, slightly tender, erythematous or some purpuric, discrete macules, papules, and patches on the both upper and lower extremities including palms and soles (Fig. 1). A skin biopsy showed an unremarkable epidermis but a dense linear and nodular infiltrate composed predominantly of neutrophils that was localized to the eccrine sweat units with slight extension to the neighboring periglandular area and subcutis (Fig. 2 and 3). Also, there was focal necrosis of eccrine coils. Apart from a mild mixed perivascular infiltrate in the middle and deep dermis, no other significant changes were observed. These clinical and histopathologic features enabled diagnosis of neutrophilic eccrine hidradenitis to be made.

One day after biopsy, chemotherapy regimen composed of cytosine arabinoside and doxorubicin was started. She tolerated well to the chemotherapy and cutaneous lesions had completely disappeared after two weeks without recurrence for follow-up period of six months.

**DISCUSSION**

Since first being described by Harris et al. in a patient with chemotherapy using cytarabine and doxorubicin for AML in 1982, approximately 50-60 cases of NEH have been reported in literatures. There is slight male predominance and mean age is 40.3 years (1-79 years). Besides only one case of NEH developed in a one-year-old baby, there has

---

**Fig. 1.** Multiple, tender, erythematous to purpuric macules, papules and patches on (A) both lower extremities and (B) forearm including palm.

**Fig. 2.** A dense linearly distributed nodular neutrophilic infiltrate around eccrine sweat units extends to the subcutis (H&E, ×40).

**Fig. 3.** Neutrophilis around the eccrine glands (H&E, ×400).
been no description about infantile NEH to our
knowledge. Our patient was a nine-month-old infant
and we think that she may be the youngest patient
with NEH in literature.

Most common is the leukemic patient receiving
systemic chemotherapy, but NEH has also been
documented in individuals undergoing chemothera-
py for a number of other types of cancer. Furthermore,
there was some reports of NEH without asso-
ciation of chemotherapy. These include the infectious
NEH (HIV, Serratia marcescens, Enterobacter cloacae,
Staphylococcus aureus), drug-induced NEH (acet-
enaminophen, G-CSF, zidovudine), and paraneo-
plastic NEH. Pierson et al. described a patient
with NEH arising several weeks prior to the onset of
fulminant AML and insisted that the NEH might
develop as a paraneoplastic phenomenon in their
case. Although our patient had suffered from AML,
because the lesions developed before chemothera-
py, we might speculate that upper respiratory infection
or medication might be a possible cause or NEH
could be a paraneoplastic phenomenon in our
case.

The cutaneous lesions of NEH are featured by
the development of infiltrative or edematous
papules or plaques, asymptomatic or painful, close to
Sweet’s syndrome. The pigmented or purpuric le-
sions have also been reported. They can develop
either in a proximal disposition involving the upper
trunk, upper limbs and the face, particularly the
periorbital areas, but distal extremities including
palms and soles are rarely involved. Moreover,
when the lesions limited to the palms and/or soles
with clinical and histopathologic resemblance
with NEH in a child without history of chemothera-
py or malignancy, the term of “idiopathic palmo-
plantar hidradenitis” is used. Since our patient had
involvement of the palms and soles similar to idio-
pathic palmpoplantar hidradenitis but also leukemic
symptoms, we believe that these features are those of
true NEH.

The definite diagnosis of NEH relies on the
histopathologic examination. The hallmark of this
condition is characterized by the association of a
degeneration of eccrine glands, and dense neu-

The pathogenesis of NEH is not yet known. Two
major hypotheses, however, are proposed in the
current literatures. One is a possible direct toxic effect
on the eccrine apparatus and this may ultimately be
shown to account for many of the chemotherapy-
related cases. The other is that NEH is a hypersensi-
tivity reaction within the spectrum of neutrophilic
dermatoses such as Sweet’s syndrome. The minor
explanation for pathogenesis of NEH include the
suggestion that neutrophilic infiltrate can be sec-
ondary to sweat gland abnormalities or a paraneo-
plastic phenomenon.

No specific treatment is required and sponta-
neous resolution of lesions of NEH without scar-
ing is always observed within a few days or weeks,
but relapse can occur in some patients receiving
the same chemotherapy regimen. Antibiotics and
nonsteroidal anti-inflammatory drugs are used for
shortening of duration and relief of pain. Dapsone
can be used for prevention of recurrent NEH in a pa-
tient receiving repeated chemotherapy.

Several aspects from our patient are worth not-
ing. First, the patient was nine-month old, the
youngest age among which has been reported. Sec-
ondly, the lesions involved the relatively uncom-
mon sites of distal extremities including palms and
soles. Finally, NEH developed before chemotherapy,
suggestive of other etiologic factors such as infec-
tion, drug, or malignancy itself.

REFERENCES

1. Harrist TJ, Fine JD, Berman RS, et al: Neutrophilic ec-
ocrine hidradenitis. A distinctive type of neutrophilic
dermatosis associated with myelogenous leukemia and
2. Bachmeyer C, Aractingi S: Neutrophilic eccrine
hidradenitis. Clinics in Dermatology 18:319-330,
2000
3. Stahr BJ, Cooper PH, Caputo RV: Idiopathic plantar
hidradenitis: a neutrophilic eccrine hidradenitis occur-
ing primarily in children. J Cutan Pathol 21:289-
296, 1994
4. Moreno A, Barnadas MA, Ravella A, DeMoragas
JM: Infectious eccrine hidradenitis in a patient un-
dergoing hemodialysis. Arch Dermatol 121:1106,
1985
5. Allegue F, Rocamora A, Martin-Gonzalez M, Alonso