Infectious Eccrine Hidradenitis Associated With Intense Sun Exposure

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Practice Points

- Neutrophilic eccrine hidradenitis is a response to nonspecific stimuli in a wide spectrum of clinical settings.
- Infectious eccrine hidradenitis should be included in the differential diagnosis in patients who develop an erythematous papular eruption after sunburn.

Infectious eccrine hidradenitis (IEH), which usually manifests as singular or multiple erythematous papules or plaques, is a rare dermatosis involving an infectious agent and histologic findings identical to that of neutrophilic eccrine hidradenitis (NEH). We report a case of IEH in a 24-year-old woman who developed a pruritic, erythematous, papular rash after a sunburn. A culture of a pustule revealed methicillin-sensitive Staphylococcus aureus. *Our patient had complete resolution of her rash within 2 weeks of starting amoxicillin and clavulanate. This case of IEH and NEH related to both intense sun exposure and infection supports the hypothesis that NEH is a response to nonspecific stimuli and may occur in many different clinical settings.*

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Case Report

A 24-year-old white woman with no remarkable medical history developed a sunburn during a record-breaking heat wave in New York, New York.[1] As the sunburn was resolving a week later, a pruritic, erythematous, papular rash appeared on her upper back. She presented with a linear distribution of erythematous papules on the flanks, grouped papules and pustules on the back, and papules on the legs in areas of resolving sunburn (Figure 1). It was her first sun exposure of the season, and the patient previously had used the same sunscreen, which she applied on all parts of her body. Her current medications included desogestrel and ethinyl estradiol, naratriptan hydrochloride, and loratadine and pseudoephedrine.

Histology showed a superficial and deep perivascular infiltrate of lymphocytes, histiocytes, and neutrophils with prominent neutrophilic infiltration of eccrine glands (Figure 2). In the overlying epidermis, there was focal parakeratosis with a collection of neutrophils and an aggregate of gram-positive cocci. Special stains for fungi and acid-fast bacilli failed to reveal microorganisms. A culture from a sample pustule revealed moderate growth of methicillin-sensitive *Staphylococcus aureus*. The patient was started on amoxicillin and clavulanate (500 mg/125 mg twice daily for 7 days) and the rash began to improve. It completely cleared within 2 weeks. Routine health maintenance examinations were normal at 1-year follow-up.

Comment

Neutrophilic eccrine hidradenitis (NEH) is a rare benign dermatosis that involves an inflammatory reaction of the eccrine glands.[2] Clinically, it usually appears as singular or multiple erythematous papules or plaques that typically are asymptomatic.[3] Pustules can occur in isolation or in erythematous plaques. The lesions may develop anywhere on the body,

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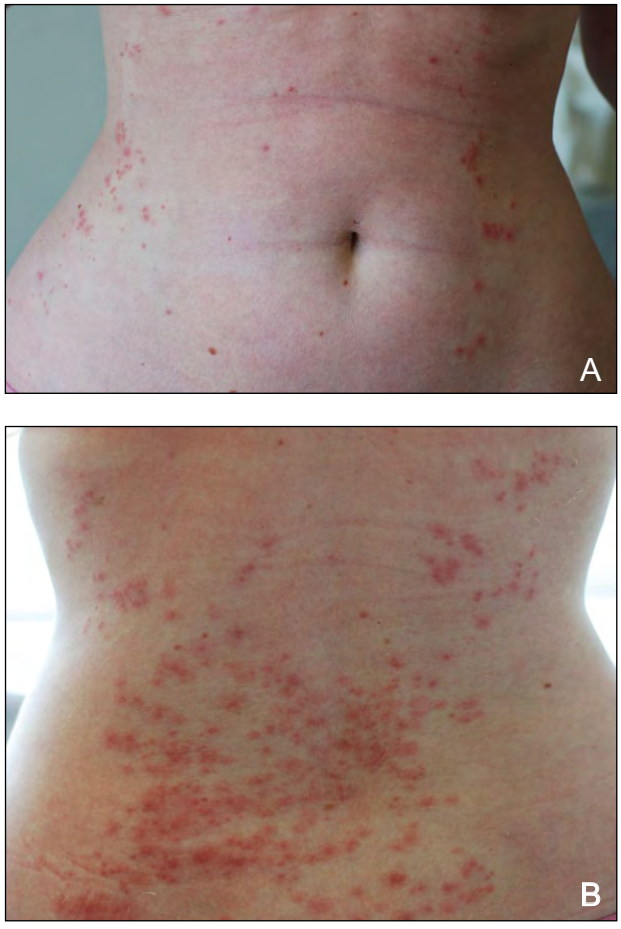

Figure 1. Linear distribution of erythematous papules on the lateral abdomen and flanks (A). Grouped papules and few pustules on the back (B).

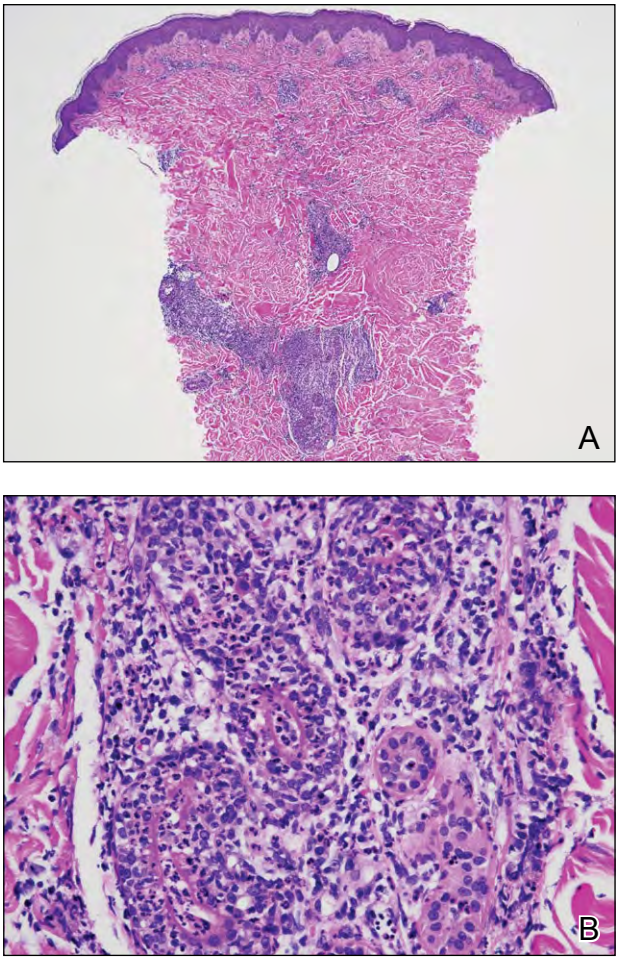

Figure 2. A punch biopsy specimen demonstrated a superficial and deep perivascular and periadnexal infiltrate of lymphocytes, histiocytes, and neutrophils (A)(H&E, original magnification ×40). Note the prominent neutrophilic infiltration of eccrine glands associated with foci of vacuolar degeneration of secretory epithelium (B)(H&E, original magnification ×400).

though the groin and axillae typically are spared. Histologic findings include a neutrophilic infiltrate around and within eccrine glands associated with necrosis of eccrine epithelium.[3]

Neutrophilic eccrine hidradenitis was initially described and is most commonly seen in patients undergoing chemotherapy for acute myelogenous leukemia but has since been described in patients with Hodgkin disease, non-Hodgkin lymphoma, chronic lymphocytic leukemia, osteosarcoma, and other solid tumors. Most patients who develop NEH received chemotherapy treatment with cytarabine and anthracyclines prior to the onset of lesions, though other chemotherapeutic agents also have been implicated.[3] However, NEH also can herald the onset of hematologic malignancies such as acute myelogenous leukemia[4] and chronic myelogenous leukemia.[5] Although 90% of NEH cases develop in patients with malignancies,[3] the disease also has been reported in patients with human immunodeficiency virus[6-8] and those taking nonchemotherapeutic drugs, including acetaminophen[9] and granulocyte colony-stimulating factor.[10,11]

Infectious agents rarely have been cultured from lesions in patients with NEH; these cases constitute what is known as infectious eccrine hidradenitis (IEH)(Table).[12-18] Although histologically identical,[13] NEH and IEH are treated differently, as NEH usually resolves spontaneously while IEH often requires treatment with antibiotics.[15] In 1985, Moreno et al[12] reported the first known case of IEH, in which *Serratia* was cultured from recurrent cutaneous lesions in a 48-year-old man with chronic renal failure who was undergoing hemodialysis. Histology of the lesions was typical of NEH. Treatment with antiseptic soaps provided no benefit; the authors did not report that antibiotics were offered as treatment.[12] The second known case of IEH was described

Cases of Infectious Eccrine Hidradenitis

Reference (Year)	Age/Sex	Associated Medical Conditions and Medications	Tissue Culture	Blood Culture	Clinical Findings	Treatment	Outcome
Moreno et al[12] (1985)	48 y/M	Glomerulonephritis; patient was on hemodialysis	*Serratia*	N/A	Erythematous 6-mm papules on upper and lower extremities and mild pruritus, recurrent each year	Antiseptic soaps	Spontaneous resolution within 3 mo but recurrent each year; antiseptic soaps provided no benefit
Allegue et al[13] (1990)	49 y/M	Flulike symptoms	*Enterobacter cloacae* from pustule	N/A	Erythematous, violaceous, 5- to 15-mm papules and pustules on upper and lower extremities and abdomen	Intramuscular amikacin (500 mg twice daily)	Lesions resolved in 1 wk without recurrence
Taira and Gerber[2] (1992)	47 y/M	Following heart transplant 4 years prior on cyclosporine	*Staphylococcus aureus*	N/A	Recurrent, 3- to 4-mm, edematous, erythematous papules on upper arm	Dicloxacillin	Lesions resolved within 5 d
Combemale et al[14] (2000)	31 y/M	Following surgery for ependymoma: febrile, urinary tract infection due to *Escherichia coli* with recurrent ependymoma and new urinary tract infection due to *Klebsiella pneumoniae*	*Serratia marcescens*	Negative	Recurrent erythematous papules on lower extremities and abdomen	Ciprofloxacin; amoxicillin and clavulanate, and ciprofloxacin; ceftriaxone and tetracyclines	Lesions in remission after each round of antibiotic therapy

Reference (Year)	Age/Sex	Associated Medical Conditions and Medications	Tissue Culture	Blood Culture	Clinical Findings	Treatment	Outcome
Antonovich et al[15] (2004)	83 y/F	COPD, mitral valve prolapse, breast and endometrial cancer, endocarditis; medications: tamoxifen, prednisone, pulmonary inhalers	*Nocardia asteroides*	*Nocardia asteroides*	Tender erythematous plaques and nodules on right lower leg	Trimethoprim-sulfamethoxazole	Lesions resolved in 3 wk
Shih et al[16] (2005)	6–14 mo/ 6 M and 4 F	None	Coagulase-negative staphylococci	N/A	Urticarial-like erythematous plaques and nodules on extremities	Oral antibiotics (not specified)	Lesions resolved within 3 wk
Oono et al[17] (2006)	71 y/F	Bullous pemphigoid while on prednisolone	Negative but Gram stain of skin biopsy showed gram-positive cocci in lumen of spiral course of affected eccrine ducts	N/A	Erythematous infiltrated papules on chest, upper back, and extremities with small pustules and mild pruritus	None	Lesions resolved in 2 wk
Takai and Matsunaga[18] (2006)	23 y/M	Streptococcal infectious endocarditis	Negative	Group A streptococci	Tender, erythematous, 1- to 3-cm papules and plaques on trunk and extremities	Antibiotics (not specified) and valvoplasty for endocarditis	Lesions resolved after operation and antibiotic injection
Current report (2010)	24 y/F	Sunburn	*Staphylococcus aureus* from pustule	N/A	Erythematous papules and pustules on trunk and lower extremities with pruritus	Amoxicillin and clavulanate	Lesions resolved in 2 wk

Abbreviations: M, male; N/A, not available; F, female; COPD, chronic obstructive pulmonary disease.

in 1990 by Allegue et al[13] who reported a case of IEH caused by *Enterobacter cloacae* in an immunocompetent 49-year-old man. The lesions resolved after 1 week of treatment with intramuscular amikacin (500 mg twice daily). A case of NEH linked to *Staphylococcus aureus* was described in 1992 in a 47-year-old man who was taking cyclosporine following a heart transplant. After 5 days of treatment with dicloxacillin, his cutaneous lesions disappeared.[2] Although each of these 3 cases of IEH were reported in men in their late 40s, subsequent reports show that the demographic of IEH patients actually is quite varied (Table).[12-18] For instance, additional reports describe cases in which *Nocardia asteroides* was cultured from a lesion in an 83-year-old woman with IEH[15] and staphylococci were cultured from skin biopsies of infants aged 6 to 14 months who were otherwise healthy.[16]

The pathogenesis of NEH and IEH is not fully understood, but various mechanisms have been proposed. Because NEH is classically associated with chemotherapy treatment, it has been postulated that it occurs due to an accumulation of cytotoxic agents in eccrine glands, leading to necrosis and activation of neutrophils.[19] An infectious etiology also is possible,[2,12-18] as microorganisms have been seen in the affected eccrine duct[15,17] and cutaneous lesions completely resolve with antibiotic treatment. It is not well understood how infection is related to IEH. Oono et al[17] found expression of epithelial antimicrobial peptides called human β-defensins in IEH lesions and suggested a need for further studies to investigate the role of these antimicrobial agents in this disease. Because of its varied clinical presentation, it has been suggested that NEH is a response to nonspecific stimuli.[2,15]

Our patient represents an unusual case of NEH and IEH associated with both intense sun exposure and *S aureus* infection. It is not clear if sun exposure, *S aureus*, or a combination of both was the cause of the lesions. Temperatures during the week that the patient acquired the sunburn were among the highest recorded in New York City, reaching 39.4°C, which exceeded the prior record set in 1999.[1] Although the intense heat may have predisposed the patient to *Staphylococcus* folliculitis resulting in IEH, an alternative hypothesis is that the intense sun exposure may have caused trauma to the eccrine glands, leading to activation of an inflammatory response. Prior authors who reported NEH in otherwise healthy pediatric patients have suggested a similar mechanism. In 2005, Shih et al[16] studied 10 infants (median age, 9.1 months) who developed NEH during the summer, though evidence of sun exposure was not present. The authors suggested that the functionally immature eccrine glands in children may be ruptured at high temperatures, causing activation of an inflammatory cascade that attracts neutrophils. Rabinowitz et al[20] and Simon et al[21] in 1995 and 1998, respectively, hypothesized that mechanical and/or thermal trauma causes damage to eccrine glands in children with idiopathic recurrent palmoplantar hidradenitis, which is a related form of NEH confined to the palms and soles in otherwise healthy children.

Conclusion

Our patient's case of NEH related to intense sun exposure and *S aureus* infection supports the hypothesis that NEH is a response to nonspecific stimuli and can occur in a wide spectrum of clinical settings.[2,15] In a patient who develops an erythematous papular eruption after a sunburn, IEH, though rare, should be included in the differential diagnosis. A culture and biopsy of the cutaneous lesions can guide the practitioner toward the appropriate treatment modality.

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