A case of neutrophilic superficial eccrine ductitis

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INTRODUCTION

Neutrophilic superficial eccrine ductitis (NSED) is a newly described disorder first reported in 2018 in a 3-year-old Japanese boy presenting with a 4-month history of erythematous papules on the trunk and upper extremities, which were refractory to topical corticosteroid therapy.[1] Here, we present a case of NSED in a previously healthy 3-year-old Caucasian boy. His skin disease was treated with therapy using oral cephalexin, suggesting a possible bacterial etiology.

Abbreviations used:

ICD: irritant contact dermatitis
NEH: neutrophilic eccrine hidradenitis
NSED: neutrophilic superficial eccrine ductitis

CASE REPORT

A 3-year-old Caucasian boy, who was otherwise healthy, presented with pruritic erythematous papules and plaques. The lesions were focused on the upper and central portions of the trunk as well as upper extremities and failed to clear with a 3-month course of topical corticosteroids. He was clinically diagnosed with scabies 3 weeks prior to the presentation, which worsened following the application of permethrin cream 5%, suggesting permethrin hypersensitivity. He then failed to improve with topical pimecrolimus cream 1%.

Physical examination revealed erythematous papules and plaques in a V-shaped distribution on the chest and back (Fig 1, *A*), with erythematous papular eruptions on the bilateral aspect of the upper portion of the arms (Fig 1, *B*) and upper portion of the thighs, sparing the palms, axillae, soles, and lower extremities. Biopsy of a lesion from the back showed a neutrophilic infiltrate within and surrounding the eccrine ducts, sparing the coiled secretory glands (Fig 2), supporting a diagnosis of NSED. The patient's lesions fully cleared with oral cephalexin (40 mg/kg/day) administered for 10 days. A month later, the patient returned with miliaria-like erythematous papular eruptions on the neck and ears, with a V-shaped distribution along the chest and back. A 14-day course of oral cephalexin (50 mg/kg/day) coupled with topical chlorhexidine soap 4% cleared the patient's lesions, and he remains clear of lesions 3 months after the completion of the therapy.

DISCUSSION

The differential diagnoses of our patient's lesions included scabies, irritant contact dermatitis (ICD), miliaria, neutrophilic eccrine hidradenitis (NEH), and psoriasis. The clinical and histopathologic characteristics of each skin disorder were compared with those observed in our case to confirm the diagnosis of NSED.

Scabies lesions are characterized by small, extremely pruritic, erythematous papules, vesicles, and/or pustules with a predilection for the hands, feet, and genitalia.[2] In infants and toddlers, nodules

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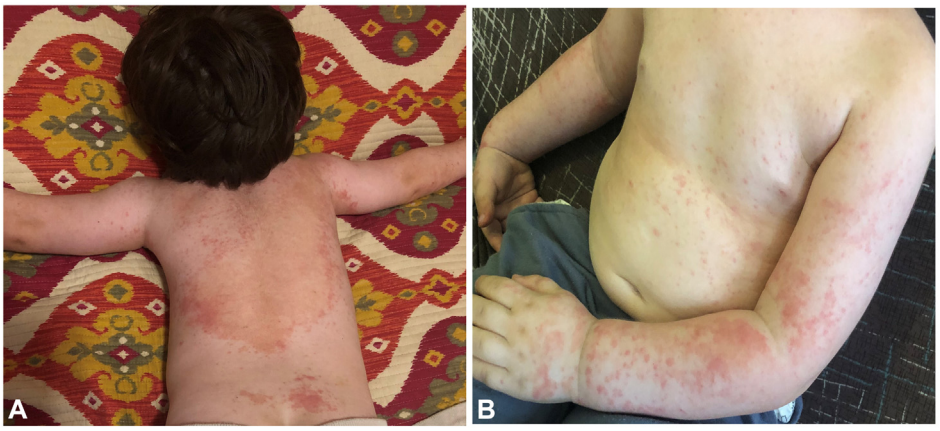

Fig 1. Clinical lesions on the patient's trunk (**A**) and upper extremity (**B**).

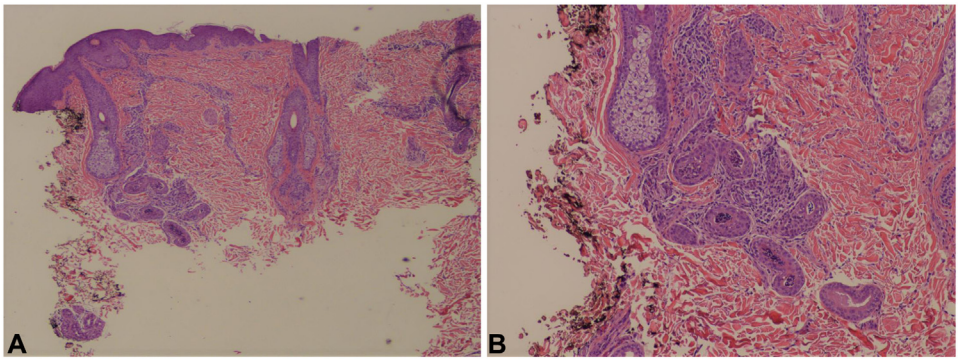

Fig 2. Low-power (×2) magnification showing inflammation of the eccrine duct along its route throughout the dermis (**A**) and higher-power (×10) magnification showing a mixed infiltrate inside and surrounding the eccrine duct; the infiltrate consists mainly of neutrophils and lymphocytes (**B**).

and eczematous lesions of the axilla or trunk can be observed.[2] Although our patient's lesions were intensely pruritic, scabies lesions typically spare the back.[2] Additionally, histopathologic findings related to scabies have shown the presence a mixed infiltrate consisting of lymphocytes, eosinophils, and histiocytes, without neutrophils, in the dermis as well as mites or eggs in skin scrapings.[3] The patient's lack of response to permethrin further provided evidence against the diagnosis of scabies.[4]

ICD in response to permethrin was suspected because of the worsening of the patient's symptoms after the application of permethrin cream 5%. ICD typically presents as erythema, edema, vesicles, bullae, and oozing at the sites of contact within minutes to hours.[5] The histopathology of ICD is characterized by a perivascular mononuclear infiltrate with intraepidermal vesicles, bullae, and necrosis of keratinocytes.[6] The patient's failure to improve with the topical corticosteroids and pimecrolimus further argued against the diagnosis of ICD.

Miliaria was also a part of the differential diagnoses. A rash of miliaria is typically papulovesicular or pustular, with background erythema, similar to our patient's erythematous papules. Although periductal inflammation in miliaria pustulosa is predominated by neutrophils,[1] the additional epidermal changes in intracorneal spongiotic vesicles that characterize miliaria were absent in our case. The sparing of the folds and prolonged disease course further argued against the diagnosis of miliaria.[1]

The lesions of NEH are painful, with erythematous edematous plaques located on the extremities, trunk, face, and palms.[7] and are unlike those of NSED, which are pruritic. The histopathology of NEH is characterized by patchy neutrophilic infiltrates surrounding the eccrine glands, with necrosis of the secretory cells and metaplasia of the sweat glands.[8] Unlike NSED, NEH undergoes spontaneous resolution within 1-2 weeks.[9] NEH is a reactive disorder and has been reported in response to

various chemotherapeutic agents, bacteria, and HIV[10,11]; however, our patient was not on any systemic therapy.[1]

Psoriasis was also considered as a differential diagnosis because our patient initially presented with pruritic erythematous plaques on the trunk and extremities with micaceous scale. The histopathology of psoriasis is characterized by neutrophils within the stratum corneum rather than surrounding or within the eccrine ducts. Additional findings related to psoriasis, including those related to epidermal hyperplasia and parakeratosis, were not present in our case.

NSED was first described in a previously healthy 3-year old Japanese boy with a 3-month history of erythematous papules on the trunk and upper extremities, which worsened with topical corticosteroid therapy.[1] We found several similarities between this and our case, including the ages of both the patients, appearance and distribution of the lesions, and refractory, persistent nature of the disease. Additionally, histopathologic examination of the lesions in the report also revealed neutrophilic infiltration within the epithelium and lumen of the sweat ducts in the lower layers of the epidermis and upper dermis and sparing of the coiled secretory glands.[1] Similar to our case, the patient in this case showed a good response to systemic antibiotics, which improved the appearance of the lesions. This suggests that systemic antibiotics are effective in the treatment of NSED and identification of a possible bacterial etiology.[1]

A third case of possible NSED was reported in Japanese language. It was the report of a 1-year-old Japanese boy with erythematous eruptions.[11] His clinical and histopathologic features were similar to those in our case; however, the patient was diagnosed with generalized NEH.[11] The refractory and prolonged courses of the lesions and absence of neutrophilic infiltration around the secretory glands, both of which are essential for the diagnosis of NEH, favored the diagnosis of NSED in this patient. A diagnosis of NSED in this patient would have increased the number of reported cases of NSED to 3.

CONCLUSION

There are just 2 reported cases of NSED, including the present case, in the English language literature. The findings consistent in both the cases are the erythematous papules on the trunk and upper extremity, prolonged disease course, histopathology revealing neutrophilic infiltration within and around the eccrine sweat ducts, sparing of the coiled secretory glands, and effectiveness of the systemic antibiotics in lesion clearance. In order to further elucidate the pathophysiology, natural history, and efficient treatments for this disorder, a larger case series is required.

Conflicts of interest

None disclosed.

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