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Letter to the Editor

A case of cholinergic urticaria with localized hypohidrosis showing sweat gland eosinophilic infiltration

Dear Editor,

Cholinergic urticaria is sometimes accompanied by idiopathic pure sudomotor failure (IPSF). The present case is the first report of cholinergic urticaria associated with localized hypohidrosis showing sweat gland eosinophilic infiltration.

A 39-year-old Japanese woman presented a 2-year history of persistent urticaria. Six months prior to her visiting to our hospital, she began to experience painful wheals during her daily bathing.

The physical findings showed punctate wheals and/or a flare response on her forearm and trunk. Neurological examinations and blood test for immunological disorders was negative. Serum immunoglobulin E level was within the normal range. The thermoregulatory sweat test showed diminished black dots on the left leg (Fig. 1a and b). Thermographic testing showed a decrease in skin temperature 30 min after exercise in all areas except the left leg (Fig. 1c). Skin biopsies taken from both the hypohidrotic and wheals areas were examined. In the wheals area, the epidermis was unchanged and significant edema was not observed in the upper dermis. Eosinophils were infiltrated around sweat glands and the blood vessels though vasculitis was denied (Fig. 2a). There was no evidence of atrophy of the sweat glands or occlusion of the duct spaces (Fig. 2b). Eosinophils were infiltration around the eccrine glands as well (Fig. 2b). In the hypohidrotic areas, eosinophils were not infiltrated around of the glands (Fig. 2c). There was no evidence of atrophy of the sweat glands or occlusion of the duct spaces (Fig. 2c). The cholinergic urticaria with hypohidrosis was treated with three sessions of methylprednisolone 1000 mg intravenously for 3 days, but it was not responsive. Cyclosporine therapy with the dose of 3 mg/kg/day then began and resulted in no recurrence of urticarial lesion with burning pain in 2 months.

The pathogenesis of acquired idiopathic generalized anhidrosis (AIGA) is poorly understood and the pathological findings vary.¹ Among the AIGA subtypes, IPSF shows a relatively high incidence. Recent reports suggest that T cell-mediated immune response,² reduced expression of the acetylcholine receptors or acetylcholinesterase in the eccrine glands^{3–5} may be involved in the pathogenesis of IPSF. The interesting point in the present case is that eosinophilic infiltration was seen in the sweat glands of the wheals lesions, while CD3-positive T cells rather than eosinophils infiltrated into the hypohidrotic areas. The patient did not present atopic dermatitis, asthma, or allergic rhinitis manifestations, and the number of eosinophils in the blood was not elevated. We

hypothesized that the infiltrated eosinophils may damage the sweat glands to induce cholinergic urticaria. The present case is the first case report of cholinergic urticaria with hypohidrosis showing sweat gland eosinophilic infiltration. However, the reason why eosinophilic infiltration was not observed around the sweat

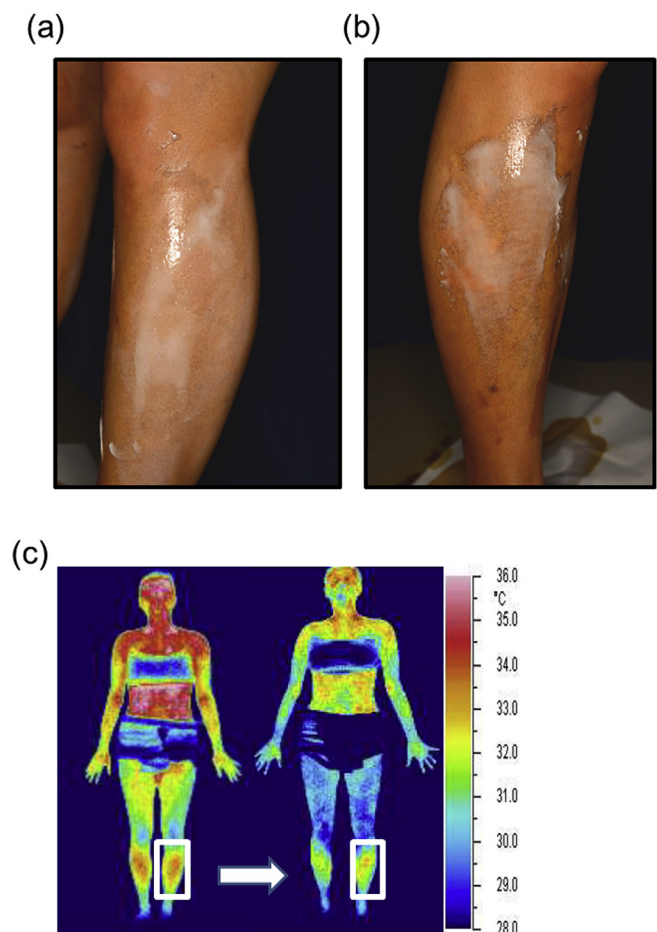
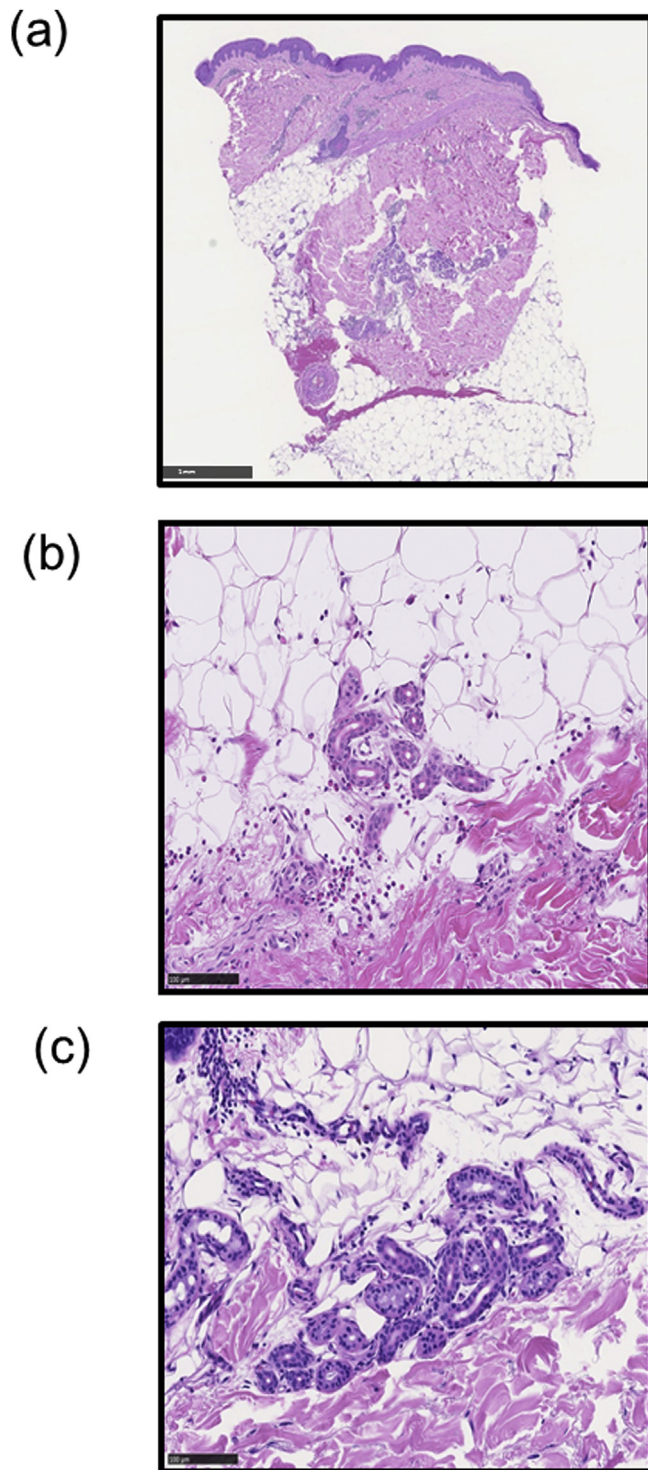


Fig. 1. Thermoregulatory sweat test using iodine-starch method and thermography. (a) Black dots were observed on the right leg. (b) No black dots were observed on the left leg. (c) Thermographical illustrations are shown just before physical exercise at 28 °C and 30 min after the exercise. The left side shows the areas before exercise and the right side shows the areas after exercise.

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glands in the hypohidrosis area, and pulse therapy was not effective for cholinergic urticaria and hypohidrosis is obscure. Careful observation have been continued to prevent developing into IPSP.

Conflict of interest

The authors have no conflict of interest to declare.

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Fig. 2. Histological observation of the wheals and hypohidrotic areas. **(a)** Wheals area. Hematoxylin and eosin staining (Scale bar: 1 mm). **(b)** Sweat glands in the wheals area (Scale bar: 100 μ m). **(c)** Sweat glands in the hypohidrotic area (Scale bar: 100 μ m).