

Correspondence

Unique case of postural cholinergic urticaria induced by a standing position

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Cholinergic urticaria (CU), which is characterized by short-lived, pinpoint weals accompanied by itch, occurs in response to physical exercise, passive warming, emotional stress, or hot food and beverages.¹ The classification of CU is categorized into four subtypes based on its cause: (i) induction by poral occlusion, (ii) acquired generalized hypohidrosis, (iii) sweat allergy and (iv) idiopathic.² However, at present the pathogenesis of CU is not completely clarified. Several clinical variations have shown that only a few weals may occur in the mild form, and that a slight increase in body temperature may induce extensive wealing accompanied by wheezing and shortness of breath in severe cases.³ Older children and young adults aged 10–30 years, are mainly affected,⁴ and many patients with mild disease do not seek medical advice. Because of the clinical variations of CU, diagnosis is not always easy. We report a case of postural CU induced by changing from a supine position to an upright standing position; this is the first such case reported, to our knowledge.

The patient was a 16-year-old Japanese high-school student, who presented with a 3-year history of thumbtip-sized pruritic and/or painful oedematous urticarial erythema on his legs, which appeared numerous times with brief disappearance and reappearance. In particular, the symptom appeared immediately after assuming a standing position and resolved within an hour. The patient did not have a history of orthostatic hypotension. The patient's attending physician had prescribed fexofenadine and diphenhydramine cream for urticaria during the previous 2 months, but despite these interventions, the patient's skin symptoms continued under the same conditions. The patient then presented to our hospital with a diagnosis of intractable idiopathic urticaria.

After admission, a provocation test was conducted (Fig. 1a). Ten minutes after the patient moved from a supine to a standing position, a number of pruritic urticarial erythematous weals were observed on both his legs (Fig. 1b).

Histopathological examination of a skin biopsy revealed mild oedema and perivascular lymphocytic infiltration

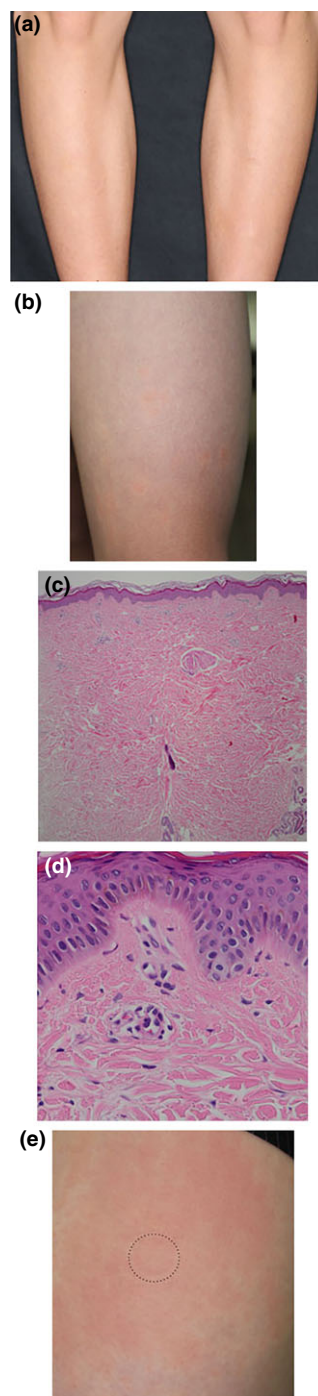


Figure 1 (a) No skin eruption on the legs occurred while the patient in a supine position, but (b) after 10 min of standing, a number of weal-like pruritic and/or painful erythematous plaques were visible on the bilateral lower limbs. (c,d) Biopsies taken after 10 min of standing revealed mild oedema and perivascular lymphocytic infiltration mainly in the upper dermis. Haematoxylin and eosin, original magnification (c) $\times 40$; (d) $\times 400$. (e) After quantitative sudomotor axon reflex test, weals appeared at the site of acetylcholine application.

mainly in the upper dermis, and these findings were compatible with urticaria (Figs 1c–d).

Furthermore, we evaluated the patient's sweat production by means of iodine–starch reaction at 30 °C and 60% humidity, and a quantitative sudomotor axon reflex test (QSART) on his forearm. For the QSART test, acetylcholine was applied to skin iontophoretically,⁵ which resulted in induction of urticaria around the site of acetylcholine application, indicating the presence of CU (Fig. 1e). No differences between the patient and a number of healthy volunteers were observed for these tests.

The remarkable nature of this case was that the trigger of urticaria on the lower leg was movement into a standing position. As symptom provocation with acetylcholine administration was also found in this case, we speculate that this effect is a physiological response to standing, from factors such as venous pressure or blood flow volume, which may induce the nerves to release acetylcholine and induce urticaria. Further examination of similar cases will afford a better understanding of the underlying mechanisms.

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Conflict of interest: the authors declare that they have no conflicts of interest.

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