

LETTER TO THE EDITOR

Elevation of serum carcinoembryonic antigen in a case of cholinergic urticaria with failed detection of hypohidrosis by the conventional starch–iodine test

Dear Editor,

Acquired idiopathic generalized anhidrosis (AIGA) is defined as non-segmental generalized hypohidrosis/anhidrosis without any dermatological or neurological causes except limited dysfunction of the autonomic nervous system only in sweating. Idiopathic pure sudomotor failure (IPSF), the most common subtype, presents distinct features, such as sudden onset in youth, absence of any specific skin lesion and favorable response to systemic corticosteroids including repeated methylprednisolone pulse therapy;^{1,2} dysfunction of acetylcholine transmission can be related to the pathomechanism.³ Recently, serum carcinoembryonic antigen (CEA) level has been proposed as a marker reflecting the disease activity of AIGA as well as a conventional starch–iodine test.^{2,4,5} Here, we report a case of cholinergic urticaria (CU), in which serum CEA elevation and lymphocytic infiltration around eccrine sweat glands were observed but the conventional test failed to detect hypohidrosis.

A 27-year-old Japanese man serving in the self-defense forces presented with sickness accompanied by a history of decreased sweating and occasional heatstroke for 7 months. In conditions usually inducing sweating, such as bathing and physical exercise, he was aware of the appearance of disseminated miliary-sized wheals on the body with itchy and prickling sensation. Blood test only showed elevated serum CEA level (19.1 ng/mL; normal, < 5.0) but no abnormality in cell counts, biochemistry, thyroid function test, immunoglobulin E and anti-SSA/B antibody. No intracranial lesion was found by magnetic resonance imaging. A modified Minor test failed to detect obvious hypohidrosis, or to induce wheals or excessive hyperthermia (Fig. 1). However, histopathology showed moderate lymphocytic infiltration around the secretory portion of eccrine sweat glands, where increased CEA expression was revealed by immunohistochemistry. These findings led to the diagnosis of CU without apparent hypohidrosis detected by the conventional Minor test. Oral olopatadine hydrochloride (10 mg/day)

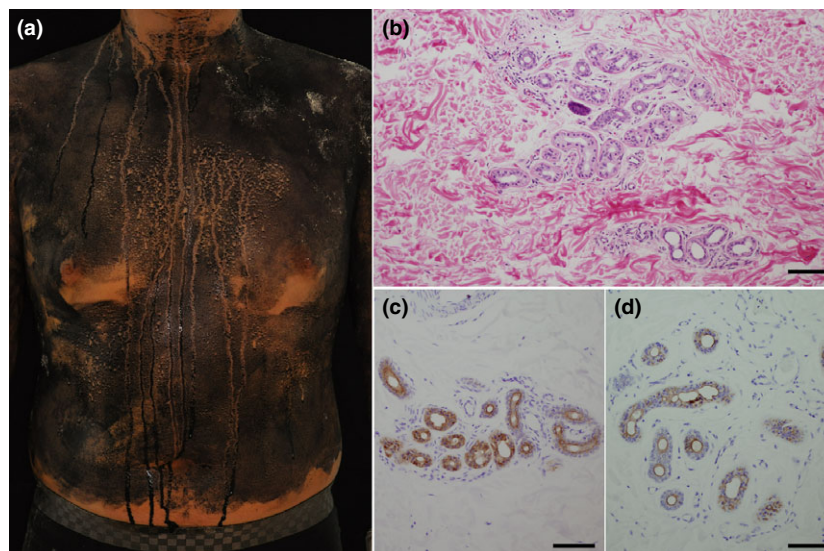


Figure 1. (a) Modified Minor test of the case. The modified Minor test is performed following the previous report.⁶ The method failed to detect the decreased sweating. (b) Histopathology showed moderate lymphocytic infiltration around the secretory portion of eccrine sweat glands. (c) Immunohistochemistry for carcinoembryonic antigen (CEA) in eccrine sweat glands of the case and (d) a healthy control. Rabbit polyclonal anti-CEA antibody (DAKO, Glostrup, Denmark) was employed for the staining. CEA expression was the case versus control. Scale bars, 100 μ m.

and repeated bathing improved the symptoms, and there was no episode of heatstroke at 6 months after the diagnosis.

At present, there is no easily repeatable and quantitative marker reflecting generalized sweating dysfunction. Serum CEA level has been proposed as the clinical marker of AIGA,^{2,4,5} and this report further suggests that it may increase even in cases of CU, in which clinically obvious hypohidrosis cannot be detected by the conventional Minor test.

For the standardized treatment, a sensitive clinical marker other than the conventional starch–iodine test is indispensable. The elevation of serum CEA level is frequently detected in patients with AIGA and IPSF.^{2,4,5} In this case, histopathology and serum CEA level suggested subclinical hypohidrosis, but the modified Minor test did not. His routinely high physical load as a self-defense forces personnel and lesser sensitivity of the starch–iodine test may be associated with the inconsistency of his subjective symptoms and the result of the conventional sweating test. Nakazato *et al.*⁵ also reported a case of partially anhidrotic CU with high serum CEA level who showed generalized anhidrosis later. Collectively, serum CEA level can increase earlier than clinical sweating dysfunction of hypohidrosis.

Carcinoembryonic antigen is a useful biomarker reflecting activity of generalized anhidrosis, and it should also be remembered that serum CEA can increase even in CU without apparent hypohidrosis detectable by the conventional sweating test.

ACKNOWLEDGMENTS: This study was supported by the Novartis Foundation.

CONFLICT OF INTEREST: None declared.

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