

Ice Cream Urticaria

To the Editor:

Cold urticaria symptoms are usually limited to cold-exposed skin areas, but the condition is potentially life-threatening.¹

CASE REPORT

In August 2009, a previously healthy 43-year-old man experienced sudden dyspnea when eating ice cream and was soon unconscious. He was treated emergently for anaphylaxis, recovered promptly, and was referred to our hospital. He had no history of similar episodes and was not taking drugs. Physical examination findings were normal. Total serum IgE was 998 IU/mL (normal <358). Other laboratory tests were within normal limits. Radioallergosorbent tests for specific IgE antibodies were all negative including egg yolk and milk. Skin prick tests were negative for egg yolk and milk. Apart from ingredients of ice cream, we suspected cold hypersensitivity. An ice cube applied to the forearm for 5 minutes resulted in a wheal on the provocation area within 3 minutes of removal, disclosing he had cold urticaria, although he had previously been unaware. Next, to compare local serum histamine concentrations before and after cold stimulation, blood samples were taken from the left hand before and 5 minutes after placing the left hand on ice. Histamine concentration increased from 0.76 ng/mL to 216 ng/mL. Finally, to confirm the anaphylaxis while eating ice cream had been due to a mucosa symptom of cold urticaria, an oral ice cube challenge test was conducted. Because cold urticaria causes edema in only cold-stimulated areas, he lay on his right side with the ice cube touching only his right buccal mucosa. Marked edema developed on his right buccal mucosa (Figure). Management of this patient started with strict avoidance of cold foods/drinks, cold water, and 3 histamine H1 blocker medications.

In December 2009, one day when diurnal temperatures were below freezing, he had sudden dyspnea upon going outside. The patient suffered more severe quality of life impairment, necessitating additional therapy. Concomitant cyclosporine 5 mg/kg daily allowed him to go out with a mask. His residence (Chitose, Japan) had an average temperature of −4 degrees Celsius and minimum temperature

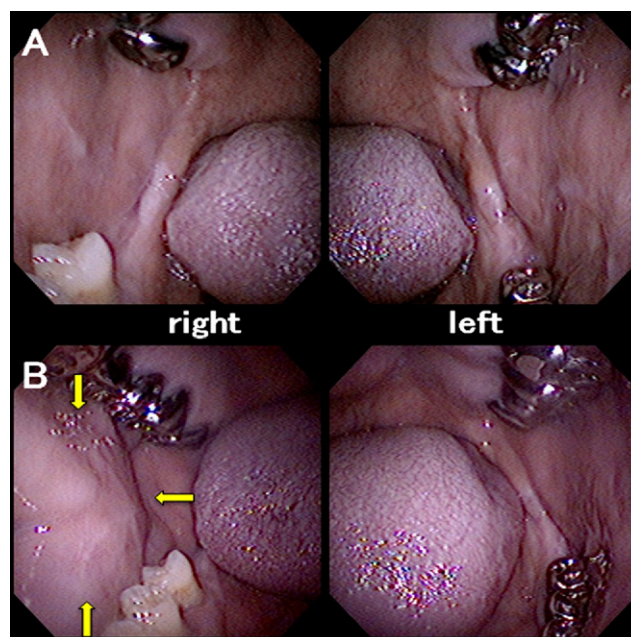


Figure Oral ice cube challenge test. The patient lies on his right side with the ice cube touching only his right side of buccal mucosa. (A) Before the test. (B) Three minutes after the ice cube provocation for 5 minutes, marked hard edema develops on his right buccal mucosa (arrow).

of −23 degrees Celsius from December 2009 to February 2010. In March 2010, as diurnal temperatures were not below freezing, cyclosporine was decreased to 3 mg/kg daily. The patient, who is seen regularly, has had no relapses of cold urticaria.

DISCUSSION

Cold urticaria can affect people of various ages, most frequently young adults. The estimated incidence is 0.05%, but varies among studies and geographical regions; incidences are higher in regions with cold climates. Half of the patients obtain remission or at least symptom improvement within 5 years.² Each patient has specific cold provocation thresholds for temperature and time.³ Although the mechanism underlying the etiology of cold urticaria remains unclear, mast cell-released histamine in the cold-exposed area is considered a principal inflammatory mediator.⁴ Avoidance of cold exposure is the best prophylaxis, and large-dose antihistamines are the first choice of medication. Further treatment options for refractory cases reportedly include corticosteroids, cyclosporine, or anti-IgE (omalizumab).²

Funding: None.

Conflict of Interest: None of the authors have any conflicts of interest associated with the work presented in this manuscript.

Authorship: All authors had access to the data and played a role in writing this manuscript.

In summary, we reported a severe case of cold urticaria whose symptom in the oral mucosa caused an anaphylaxis. To our knowledge, we first demonstrated it by an oral ice cube test. In a case of the patient living in a region with cold climate, more strict managements for cold hypersensitivity are needed. Our patients responded well to cyclosporine.

Mitsuhiro Ota, MD, PhD^a
Hiroyuki Kawasaki, MD, PhD^a
Masashi Horimoto, MD, PhD^b
Departments of ^aDermatology and
^bInternal Medicine
Chitose City Hospital
Chitose, Japan

doi:10.1016/j.amjmed.2010.04.034

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