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URTICARIA
Interesting Cases

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Case 4

Exercise-induced Angioedema

Rungsima Kiratiwongwan, Papapit Tuchinda,
Leena Chularojanamontri, Kanokvalai Kulthanan

CASE REPORT

A 36-year-old Thai female presented at the Siriraj Urticaria Clinic, Department of Dermatology, Faculty of Medicine, Siriraj Hospital, Mahidol University, Bangkok, Thailand with history of recurrent urticarial wheals and sporadic mild angioedema on both eyelids for 1 year. These lesions were not associated with any predisposing factors or physical stimuli. She also complained of having isolated angioedema on her extremities after exercise for 3 years. The swelling developed approximately one hour after exercise, and it usually persisted for a duration ranging from 4 hours to 14 hours. This acral angioedema was never accompanied by systemic symptoms, such as shortness of breath or dyspnea. Moreover, these symptoms were not associated with the intake of any specific foods or medications.

This patient's only comorbidity at the time of this report was allergic rhinitis. She used to take antihistamines to relieve her allergic rhinitis symptoms as needed, and denied any other habitual medication intake. No other family members had history of urticaria or angioedema.

All components of the physical examination were normal. Provocation test was performed by exercise on a stationary bicycle. After 15 minutes of bicycling, her pulse rate increased to 108 beats per minute and asymptomatic acral edema developed on both hands and feet with no evidence of wheals or angioedema elsewhere **(Figs. 1A and B)**.

Her blood pressure and other physical examination, including cardiovascular and respiratory examination, were unremarkable.

Laboratory tests conducted included complete blood count, erythrocyte sedimentary rate, blood chemistry, serum immunoglobulin E levels, antithyroglobulin, antithyroid peroxidase, anti-nuclear antibodies, plasma d-dimer level, and urinalysis. All of these investigations were within normal limits. Autologous serum skin test was negative. Skin prick test showed positivity to house dust mite, which clinically correlated with the patient's

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Figs. 1A and B: Hands (A) and feet (B) after bicycling with a stationary bicycle for 15 minutes.

allergic rhinitis symptoms. The patient was diagnosed as a case of exercise-induced angioedema and chronic spontaneous urticaria with angioedema.

The patient was prescribed desloratadine 5 mg/day, which completely controlled her urticarial rashes and acral angioedema. Repeated bicycling provocation test 6 months later showed no swelling of hands or feet.

DISCUSSION

Angioedema is a sudden nonpitting, skin-colored or sometimes erythematous swelling of skin, mucosal membrane, or both,[1-3] and it may coexist with urticaria.[1,2,4] Several causes of angioedema have been identified, including exercise-induced angioedema.[1-3] To our knowledge, exercise-induced angioedema at the acral parts of the body has not been reported.

We concluded that the cause of acral angioedema in this patient was exercise since she had positive provocation test with a stationary bicycle, which was static and did not cause vibratory stimuli. Exercise-induced angioedema should be differentiated from vibratory angioedema, which could be caused by bicycling on rough ground.[5]

Angioedema could be associated with physical stimuli, such as cold stimuli and increased core body temperature.[6] The prevalence of exercise-induced angioedema among patients with angioedema without wheals at an allergy and immunology center in Argentina was 0.7% (2 in 280 cases).[2] The pathophysiology of urticaria and angioedema triggered by physical stimuli is not clearly understood, but it is thought to be related to mast cell stimulation that result in the release of several vasoactive mediators, such as histamine.[7] Sweat allergy and preexistence of eczema in atopic dermatitis patients leading to sweat leakage have also been purposed to play a role in the pathogenesis of peripheral angioedema from cholinergic urticaria.[3,8]

Kato T, et al.[4] reported a 34-year-old Japanese patient with angioedema at her eyelids without urticaria after exercise. Plasma histamine concentration level measured after her provocation test remained unchanged. In contrast, that same group assessed plasma histamine concentration in another patient who had cholinergic urticaria without angioedema after

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exercise challenge, and they found the plasma histamine level to be elevated. Accordingly, they suggested that the pathogenesis of cholinergic urticaria and exercise-induced angioedema might be different.

Washio K, et al.[3] reported 15 cases of cholinergic urticaria with palpebral angioedema. All patients were female and had hypersensitivity to autologous sweat (positive reaction against an autologous sweat skin test). Almost all of those patients had atopy. Patients with atopic dermatitis often exhibited wheals on eczematous lesions since sweat might easily leak in these lesions.

CONCLUSION

This is a case of an exercise-induced angioedema with acral edema and coexisting chronic spontaneous urticaria. The physical stimulus that caused her angioedema could be identified, but her urticaria was spontaneous. Hence, the causes of angioedema and urticaria in this patient might be different. The pathophysiology of exercise-induced angioedema requires further study.

KEY MESSAGES

- Patient of angioedema may present with acral edema.
- Acral angioedema may not be accompanied by systemic symptoms.
- Causes of angioedema and urticaria in the same patient may be different.

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