Case Report

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A Rare Case of Pediatric Bullous Spontaneous Acute Urticaria

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Significance of the Study

- This is an extremely rare and unusual presentation of spontaneous urticaria. The patient responded quickly to antihistamine and systemic steroid treatment, and the lesions did not recur.

Keywords
Acute urticaria · Bullae · Antihistamine

Abstract

Objective: Acute spontaneous bullous urticaria is an extremely rare entity, and there are few reports with blister formation in acute urticaria patients. ***Clinical Presentation and Intervention:*** We present a 2-year-old girl who was admitted for bullous spontaneous acute urticaria; the underlying reason for this was not detected. Nikolsky's sign and Darier's sign were negative. Lesions were not compatible with erythema multiforme. However, biopsy was not allowed to be performed. Because of this, the underlying pathogenesis could not be clarified. The patient recovered by a short course of antihistamine and systemic steroid treatment, and the lesions did not recur during a 2-year follow-up. ***Conclusion:*** Short-term systemic steroid in addition to oral antihistamines resulted in prompt recovery in a patient with acute urticaria complicated by bullae.

Introduction

Bullous urticaria is a rare type of urticaria, and the number of cases is limited. Fluid-filled blisters or bullae are mostly seen in insect bites, physical urticaria such as delayed pressure urticaria, and dermographism [1, 2]. First reports related to bullous urticaria emerged in the 1950s [3, 4]. Although the exact etiology of bullous spontaneous acute urticaria is unclear, eosinophilic cells demonstrated in the dermis may be one of the triggering factors of bullous urticaria [3, 4]. Herein, we report a 2-year-old girl diagnosed with this rare type of urticaria.

Case Report

A 2-year-old girl was admitted to our department on the second day of her urticaria after the appearance of bullous lesions on her legs (Fig. 1). Urticarial lesions faded in less than 24 h, and flared up elsewhere later. Bullae were mostly seen on the lower extremities, were 1–2 cm in size, and tended to coalescence. The palms, soles, mucosae, eyes, and anal region were spared of bullous le-

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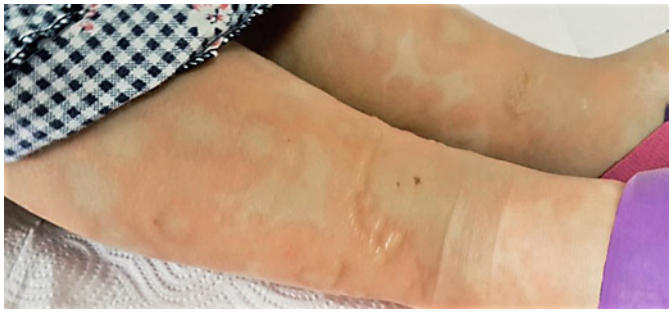

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Fig. 1. Bullous lesions at the lower extremities of the patient.

sions. In addition, Nikolsky's sign, bulla spread, and Darier's sign were all negative. The lesions were not compatible with erythema multiforme. Whole blood count, erythrocyte sedimentation rate, C-reactive protein, urine analysis, and hepatic and renal function tests were within the normal ranges; total serum IgE was 89 kU/L, and there was no eosinophilia. Serological tests for Epstein-Barr virus, cytomegalovirus, herpes simplex type 1-2, mycoplasma pneumonia, and streptococci tests were all negative. There was no history of recent drug intake or newly introduced foods. Oral hydroxyzine treatment was initiated on the day of admission, but no change was observed in the bullae or urticaria. Therefore, systemic steroid treatment (1 mg/kg/day, per os) was given on the third day of the antihistamine treatment for 3 days. The patient fully recovered at the end of a week after scabbing over the bullae with yellowish-brownish crusts on day 5. Skin prick tests with common aeroallergens and food allergens were performed 6 weeks following the disappearance of the lesions. Urticaria and bullae did not recur during a 2-year follow-up period.

Discussion

Urticaria occurs quite often in childhood especially after infections; however, associated bullae with wheals are extremely rare. Distinct from classical hives, the urticarial lesions in our patient were accompanied by blisters. Patients with bullous urticaria must be differentiated from patients with dermatitis herpetiformis, bullous pemphigoid, and bullous erythema multiforme. In dermatitis herpetiformis, lesions resemble herpetic lesions, and occur in a chronical relapsing course due to gluten hypersensitivity [5]. Bullous pemphigoid is an autoimmune disorder of the skin and mucosal membranes with chronic progression [6]. Bullous erythema multiforme is defined by classical target lesions with central blisters, which are characteristic of this disorder [7]. We differentiated our patient from all these disorders by her clinical course and the appearance of the lesions. In addition, some patients with bullous urticaria have delayed pressure, dermographism, food allergy, and insect bites [1–4, 8]. Our report differed from previous cases with regard to the lack of recurrence and an underlying cause. To the best of our knowledge, few cases of bullous spontaneous acute urticaria have been reported [4]. In the literature, infiltration of eosinophils, lymphoid cells at the base of bullae, and edema of the cutis were identified in the biopsy specimen [4]. It may be considered as an eosionophil-mediated disorder with excess cytokine release that causes blister formation. Unfortunately, a skin biopsy was not performed, because the parents did not allow the procedure. Other similar cases have been reported as repeated or chronic [1, 2]. Initially, we expected that the lesions of the patient would progress or would recur based on reports in the literature. However, lesions faded in a short time and did not recur. Although oral antihistamines are recommended as first-line treatment and short-term oral corticosteroid therapies are recommended in patients responding poorly to antihistamines [9], we administered a brief course of oral corticosteroid therapy as the bullous lesions did not fade after oral antihistamine therapy. After the treatment with systemic steroid, the blistering lesions disappeared promptly.

Acute urticaria is often a self-limiting skin disorder. Rarely, it may be accompanied by blistering hives. Similar cases are reported to recur and become chronic. Our patient differs from previous cases in terms of the benign course and rapid response to treatment. However, biopsy could not be performed and thus, the etiopathogenesis of the lesions could not be investigated.

In conclusion, acute urticaria rarely presents with bullous formation. A short course of systemic steroid treatment as well as oral antihistamines resulted in a quick recovery in the current patient who had acute urticaria associated with bullae.

Statement of Ethics

The authors have no ethical conflicts to disclose.

Disclosure Statement

The authors have no conflicts of interest to declare.

Author Contributions

P.G.Ç. followed the patient under the supervision of Ü.M.Ş. Ö.U.S. and B.E.Ş. contributed to the discussion of the case.

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