

## LETTERS TO THE EDITOR

## A non-responsive chronic autoimmune urticaria in a 12-year-old autistic girl treated with cyclosporin

### Editor

Urticaria is a heterogeneous group of skin disorders common in childhood and characterized by raised and erythematous wheals, accompanied by intense pruritus, whose aetiology often remains unknown.<sup>1</sup>

About 30% of children with chronic urticaria (CU) have an autoimmune aetiology with positive autologous serum skin test (ASST).<sup>2</sup> Moreover, an association between childhood, CU and other autoimmunity disorders such as Hashimoto's thyroiditis has been postulated: about 4% of children suffering from CU have positive anti-thyroid antibodies, although the majority is euthyroid.<sup>3</sup>

In literature, an association between CU and celiac disease, with significant clinical improvement of both disorders by a gluten-free diet, has been reported.<sup>4</sup>

CU in childhood is a self-limiting dermatosis; approximately 25% of children will achieve remission within the first 3 years from the onset of symptoms.<sup>5</sup>

As concerns therapy, every identifiable factor should be eradicated or avoided firstly; stress management strategies and treatment of underlying infections may help to control symptoms.<sup>5</sup>

Antihistamines, in particular H1 antihistamines, represent the gold standard treatment for children with CU.

Short-term use of oral corticosteroids may be required to gain control in children poorly responsive to maximal doses of H1 antihistamines.<sup>6</sup>

In children with positive ASST, the best evidence exists for the use of leukotriene receptor antagonists (LTRA, in particular montelukast) or cyclosporine A.<sup>7</sup>

Cyclosporine A is of proven safety in children; in recent years, it was introduced for the treatment of several inflammatory diseases, such as psoriasis, lupus nephritis, autoimmune hepatitis, haematological disorders and for preventing the rejection of post-transplants,<sup>8,9</sup> but no data have been reported in literature on its use in chronic autoimmune urticaria in childhood.

A 12-year-old autistic girl was referred to our Dermatological Department for clinical evaluation of a dermatosis characterized by circumscribed, slightly elevated, erythematous oedematous



**Figure 1** 12-year-old autistic with slightly elevated, erythematous oedematous papules all over the body.

papules all over the body, accompanied by intense pruritus (Fig. 1). The initial evaluation suggested the diagnosis of acute urticaria and routine serology was negative, apart from C-reactive protein.

Patient did not respond to oral intake of Desloratadine 5 mg per day; thus, she was shifted towards Hydroxyzine hydrochloride syrup 10 mL three times a day and Idrocortysone 25 mg for 5 days and 12.5 mg for another 5 days.

After 10 days of treatment, cutaneous lesions completely cleared, and only a moderate itching persisted; thus, antihistaminic monotherapy (Levocetirizine 5 mg per day) was continued for 1 month, with rapid relapse of cutaneous lesions and worsening of pruritus.

Patient failed to respond also to systemic administration of LTRA (Montelukast 5 mg per day).

Since therapy based on systemic antihistamines, antileukotrienes, and corticosteroids had no satisfying effects, an ASST was made,

and its positive result led us to the final diagnosis of autoimmune CU unresponsive to conventional therapy.

Cyclosporine therapy was started at the dosage of 4 mg/kg/day for 2 months decreased to 0.3 mg/kg/day every 2 months for 1 year of therapy, with systematic control of renal function, monthly.

In our case, Cyclosporine A permitted to gain the control of the disease without any significant side effects; at the moment, the patient is at the 14th month of consecutive therapy and she doesn't present clinical lesions and itching.

In conclusion, Cyclosporine A seems to be effective in inducing and in maintaining remission of chronic autoimmune urticaria in childhood; however, it should be emphasized that chronic autoimmune urticaria in children is yet an unlicensed indication for Cyclosporine,<sup>10</sup> and further controlled trials are needed to evaluate its side effects in long-term use and relapses after withdrawal.

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