Chronic urticaria and angioedema associated with Hashimoto’s thyroiditis in a child: A case report

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ABSTRACT

Introduction: The association between chronic urticaria and Hashimoto’s thyroiditis has been rarely reported in children. Case Report: We are reporting a case of an eight-year-old girl with chronic urticaria unresponsive to antihistaminic therapy, who was subsequently diagnosed to have Hashimoto’s thyroiditis. Patient’s urticarial lesion remitted after treatment with levothyroxine. Conclusion: We emphasize to consider testing for Hashimoto’s thyroiditis by thyroid autoantibodies and thyroid profile in cases of chronic urticaria, and starting the patient on levothyroxine for the symptomatic improvement of both chronic urticaria and Hashimoto’s thyroiditis.

Keywords: Angioedema, Chronic urticaria, Hashimoto’s thyroiditis, Levothyroxine

INTRODUCTION

Hashimoto’s thyroiditis is an organ specific autoimmune disease characterized histologically by lymphocytic infiltration of the thyroid gland. This disorder is 2–4 times more frequent in girls than in boys. Most of the affected children are euthyroid and asymptomatic. The most common clinical manifestations are goiter and growth retardation secondary to hypothyroidism. Thyroid anti peroxidase antibody (TPO Abs) and antithyroglobulin antibodies are present in 90% of the affected children [1].

Childhood chronic urticaria is a common disorder characterized by the appearance of hives for more than six weeks [2, 3]. The pathogenesis of chronic urticaria is poorly understood and the cause is unknown in majority of cases [4]. A subset of patients with chronic urticaria has been classified as autoimmune on the basis of association with thyroid autoimmunity and anti-IgE and/or anti-IgE receptor antibodies [5].

As per literature review, an association has been found between Hashimoto’s thyroiditis and chronic urticaria in adults [6], but there are only a few cases reported in children [7]. Hence, we are presenting a case of an eight-year-old girl with chronic urticaria and Hashimoto’s thyroiditis who became asymptomatic after treatment with levothyroxine.

CASE REPORT

An eight-year-old Hispanic girl referred by immunologist to our pediatric endocrinology clinic in month of September 2010 with abnormal thyroid function test and chronic urticaria. On August 2010,
A patient was presented to an immunology clinic with a two-month history of recurrent urticarial lesions over face, trunk, and extremities accompanied by swelling of lips and tongue which were non-responsive to antihistamine treatment. The patient also complained of weakness, fatigue, and significant weight gain over the period of six months. In the immunology clinic, routine laboratory analysis, allergy tests, and thyroid function tests including thyroid autoantibodies were performed. The patient was found to have high TSH (9.8 mIU/mL [< 4.6 mIU/mL]), low free T4 (0.5 ng/dL [normal limit 0.7–1.5 ng/dL]), high anti-TPO antibodies (352 IU/mL), and positive anti-Fc epsilon Receptor (anti-FCER) antibody. Routine laboratory analysis and total serum IgE were within normal limits. Allergic testing to common food allergens was negative.

On examination in the pediatric endocrinology clinic, the patient's vital signs were normal, weight 46.03 kg (>95 percentile), height 133.5 cm (75–90 percentile), and BMI- 25.83 (>97 percentile). Her physical examination showed multiple urticarial lesions over the face, trunk, and extremities, and the thyroid gland was not palpable. Systemic examination was unremarkable. The patient was started on levothyroxine 100 µg/day and antihistamine as needed for urticaria and angioedema. The patient was followed-up in February 2011; urticarial lesions were completely remitted during the visit while on levothyroxine with normal thyroid function. The patient was continued to follow-up at regular intervals in our endocrinology clinic and remained asymptomatic after one year on hormone replacement therapy with normal thyroid function test (Table 1).

**Table 1: Changes in thyroid function test with treatment**

| Thyroid Function Tests | Prior to levothyroxine treatment | 4 months after treatment | 12 months after treatment |
|------------------------|----------------------------------|--------------------------|--------------------------|
| TSH (mIU/mL)           | 9.8                              | 2.16                     | 4.15                     |
| Free T4 (ng/dL)        | 0.5                              | 1.7                      | 1.6                      |
| Anti-TPO antibodies (IU/mL) | 352                          | -                        | -                        |

**DISCUSSION**

Chronic urticaria is a common clinical condition whose etiology in about 75% of cases is unknown. A link between chronic urticaria and autoimmune thyroid diseases such as Hashimoto's thyroiditis has been proposed and studied [8]. Approximately one-fourth of chronic urticaria patients have serological evidence of thyroid autoimmunity suggesting association between them. There are various hypotheses on how these two entities might be related:

(a) Immune complexes produced in the course of Hashimoto's thyroiditis are trapped in the skin and may cause urticaria.
(b) Inflammatory cells activated in the thyroid are directed toward a cross-reactive antigen existing in the skin.
(c) There is no direct relationship between the inflammation in the thyroid and the skin, but autoimmunity directed toward FeER coexists with autoimmunity directed toward the thyroid gland in susceptible patients.
(d) There is no relationship between chronic urticaria and Hashimoto's thyroiditis, but they are both common disorders coexisting in a small percentage of patients [9].

Hashimoto’s thyroiditis with chronic urticaria in children is rare with only a few reported cases. A study by Levy et al. described eight female patients in the age group of 7–17 years with chronic urticaria and positive thyroid autoantibodies. Two of the eight patients were hypothyroid and were started on thyroxine without any improvement in urticaria [7]. In our patient, the common causes of chronic urticaria like allergy to external agents, hereditary angioedema, and occult infections were excluded and the patient was diagnosed to have Hashimoto’s thyroiditis based on high TPO antibodies titers and abnormal thyroid profile. The patient was started on levothyroxine therapy. After four months of treatment with levothyroxine, urticarial lesions disappeared without any recurrences noticed over the one year follow-up. This was in contrast with the two hypothyroid patients studied by Levy et al. whose urticaria did not respond to thyroxine treatment.

Therefore, we assume an association between chronic urticaria and Hashimoto’s thyroiditis with clinical remission of resistant urticarial lesions after levothyroxine treatment.

**CONCLUSION**

We recommend considering testing for Hashimoto’s thyroiditis by thyroid autoantibodies and thyroid profile in cases of chronic urticaria and starting levothyroxine in a hypothyroid patient with Hashimoto’s thyroiditis for its symptomatic improvement as well as for ameliorating chronic urticaria.

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**Author Contributions**
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Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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REFERENCES
1. Kliegman RM, Stanton BF, Schor NF, St.Geme III JW, Behrman RE. Nelson Textbook of Pediatrics, 19th Ed. Philadelphia: Elsevier Saunders, 2011, p2181.
2. Greaves MW. Chronic urticaria in childhood. Allergy 2000;55(4):309–20.
3. Joint Task Force on Practice Parameters. The diagnosis and management of urticaria: a practice parameter. Part I: Acute urticaria/angioedema. Part II: Chronic urticaria/angioedema. Ann Allergy Asthma Immunol 2000;85:521–44.
4. Bangash SA, Bahna SL. Resolution of chronic urticaria and angioedema with thyroxine. Allergy Asthma Proc 2005;26(5):415–7.
5. Rottem M. Chronic urticaria and autoimmune thyroid disease: is there a link? Autoimmun Rev 2003;2(2):69–72.
6. Irani C, Janmal M, Asmar G, Haji H, Halaby G. Chronic urticaria and autoimmune thyroiditis. J Med Liban 2012;60(2):88–90.
7. Levy Y, Segal N, Weintrob N, Danon YL. Chronic urticaria: association with thyroid autoimmunity. Arch Dis Child 2003;88(6):517–9.
8. Bagnasco M, Miniculiuo PL, Saraceno GS, Gangemi S, Benvenega S. Urticaria and thyroid autoimmunity. Thyroid 2011;21(4):401–10.
9. Kandeel AA, Zeid M, Helm T, Lillie MA, Donahue E, Ambrus JL Jr. Evaluation of Chronic Urticaria in Patients with Hashimoto Thyroiditis. J Clin Immunol 2001;21(5):335–47.