A VERY SEVERE DERMOPATHY AND OPHTHALMOPATHY IN PATIENTS WITH AUTOIMMUNE THYROID DISEASE: CASE REPORT AND LITERATURE REVIEW

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SUMMARY

Although ophthalmopathy (most commonly) and dermopathy (uncommon) are the extrathyroidal manifestations of Graves' disease, they are extremely rare in Hashimoto's thyroiditis. We present a case of Hashimoto's hypothyroidism with ophthalmopathy and dermopathy (the elephantiasic form of pretibial myxedema) in a 46-year-oid man. He was diagnosed as having Hashimoto's hypothyroidism with decreased thyroid hormone levels and elevated TSH, antithyroid peroxidase and antithyroglobulin antibodjes levels, and on palpation the thyroid gland was typical for Hashimoto's thyroiditis. The patient had infiltrative ophthalmopathy and elephantiatic form of pretibial myxedema. Thyroid ultrasound revealed that the parenchyma was heterogenous. Urinary glycosaminoglycans level was increased. Orbital CT scanning showed that extraocular muscles were thickened indicating the infiltrative opthalmopathy. This case demonstrates the uncommon extrathyroidal manifestation (a very severe dermopathy and ophthalmopathy) of autoimmune thyroid disease.

Key words: Autoimmune thyroiditis, ophthalmopathy, dermopathy

INTRODUCTION

The ophthalmopathy is most commonly associated (80%) with Graves' disease, it can be seen in 10% to 15% of Hashimoto's thyroiditis [1]. Besides this finding, pretibial myxedema is a late and an uncommon (10% to 12%) manifestation of Graves' disease (GD) with ophthalmopathy. However both manifestations are very rare only in Hashimoto's thyroiditis [2,3]. The very rare association between extrathyroidal manifestations of autoimmune thyroid disease and Hashimoto's thyroiditis is discussed.

CASE REPORT

In 2002, a 45 year-old-man was admitted with the complaints of weight gain, facial puffines, and prominence of eye balls continuing for about few months. At that time, he had thyroid opthalmopathy (grade 3) and pretibial myxedema (nonpitting edema accompanied by typical color changes and plaque of the skin , Figure 1), but he was clinically euthyroid. There were no symptoms of hyperthyroidism in the patient's history. Serum thyroid-stimulating hormone (TSH) level was normal (1.1 mU/l, reference range 0.4-4.0), and thyrotropin receptor antibodies (TSH-RAb) were elevated (>405 U/l, reference range 9-14). He has skipped his controls for a year. When he was reexamined, the patient was hypothyroid. He had worsening of his voice and the skin over his tibial areas was changed. He had gained more weight, had cold intolerance and progression of ophthalmopathy. The patient was clinically hypothyroid, and he had severe thyroid ophthalmopathy (Figure 2). Physical examination revealed a pulse of 78/min regular, blood pressure of 130/90 mmHg. There was no appreciable thyromegaly or nodularity in the thyroid gland. Thyroid ultrasound revealed that parenchyma was heterogenous.

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Figure 1. Bilateral erythematous infiltrative plaques in the pretibial areas of the lower extremity and foot, at the first admission.

Eye examination showed bilateral proptosis measuring 32 mm Hertel. Lateral eyeball movement was restricted and covergence was markedly affected (grade 4 ophthalmopathy). He had elephantiatic form of dermopathy at lower extremities (Figure 3). A complete blood celi count showed hypochromic microcytic anaemia with normal blood iron and total iron-binding capacity. Thalassemia minor was present which was proved by peripheric blood smear and hemoglobin electrophoresis. Serum triiodothyronine (T_3), thyroxine (T_4) and free T_4 levels were decreased with elevated TSH level (0.4 nmol/l, reference range 0.8-2.0; 44 nmol/l, reference range 64-142; 6.8 pmol/l, reference range 12-22; 58 mU/l , reference range 0.2-4.5, respectively). TSH-R, antithyroid peroxidase and antithyroglobulin antibodies were elevated (>405 U/l, reference range 9-14; 3000 U/ml, reference range 0-35l; 1870 U/ml, reference range < 60, and respectively). Urinary glycosaminoglycans level was increased (4,29 mg/ml creatinin daily, reference range 0.6-2.6). Computed tomography scan of orbit revealed marked extraocular muscle thickening (Figure 4). On the basis of these findings, hypothyroidism with ophthalmopathy and dermopathy was diagnosed, and replacement therapy with T_4 was started. The patient was discharged after 20

Figure 2. Photograph of the patient showing changes of infiltrative ophthalmopathy

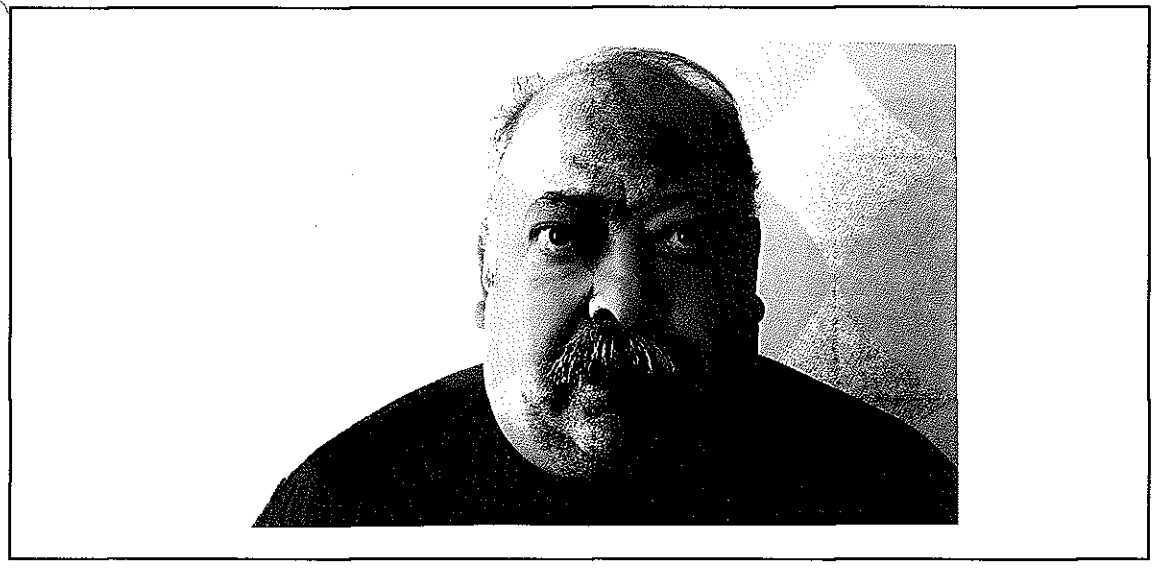
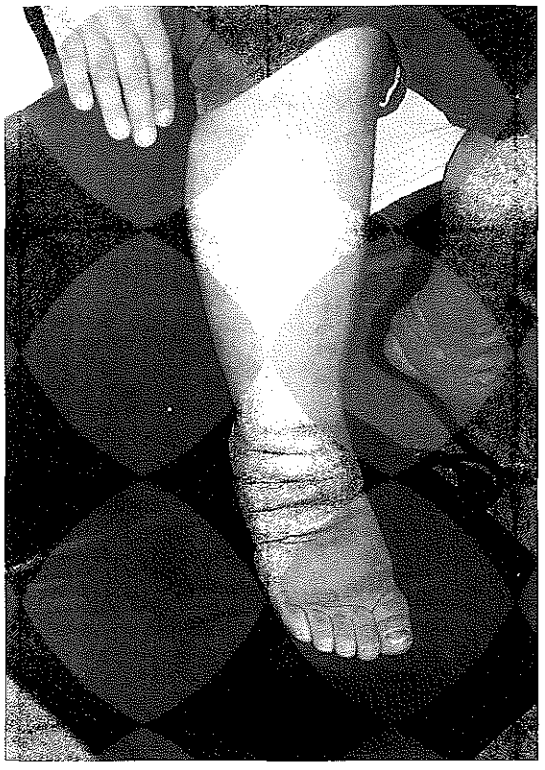


Figure 3. The elephantiasic form of PTM, the lesions were coalesce to give the entire extremity an enlarged, verruciform appearance.

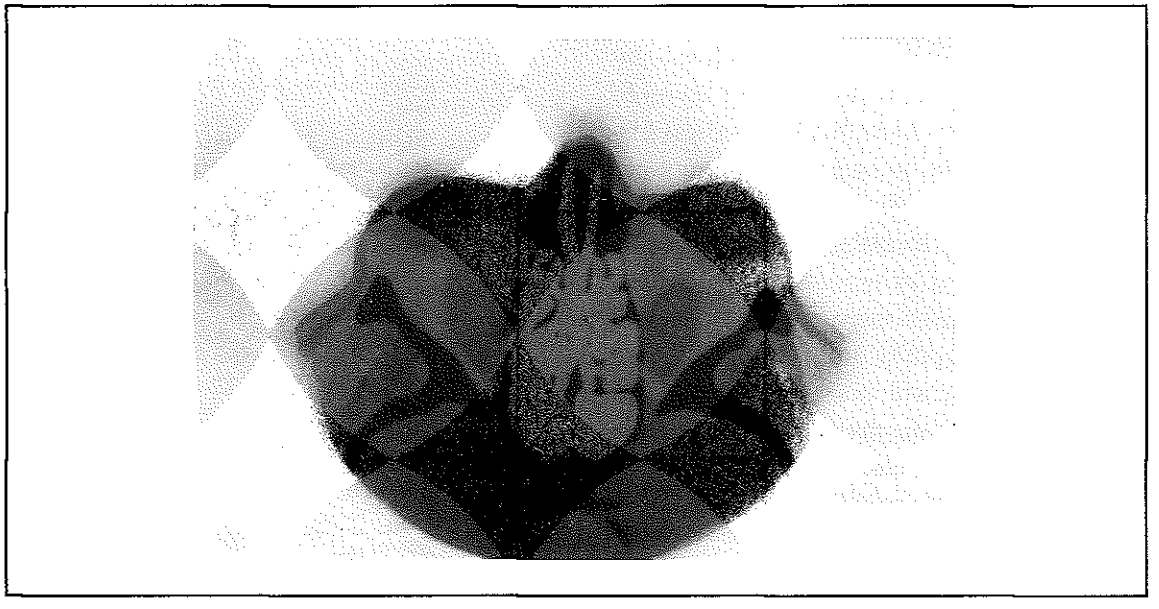


days under T_4 replacement (200 µg daily). We decided that ophthalmopathy and dermopathy treatments modalities will be considered after euthyroidism was achieved.

DISCUSSION

Autoimmune thyroid disease encompasses a spectrum of thyroid disorders with GD at one end and Hashimoto's thyroiditis at the other end [4]. Although GO is mostly associated with hyperthyroidism, ophthalmopathy may less frequently occur also in patients with noncurrent or past evidence of thyroid hyper- or hypofunction [5,6]. We here report a case of Hashimoto's hypothyroidism with ophthalmopathy. Brownlie et al.[7] reported that the prevalence of ophthalmopathy associated with Hashimoto's thyroiditis was only 2%, which is a very rare association than with GD. About 15% of patients with severe Graves' ophthalmopahy havc the cutaneous manifestation, and 0.5-4.3% of patients with a history of thyrotoxicosis have thyroid dermopathy has been reported [5,8-10]. In the literature, only two case reports of Hashimoto's thyroiditis with ophthalmopahy and dermopathy have been reported [11,12]. On the first admission, the patient had significant eye disease and dermopathy. The milder form of dermopathy had progressed to elephantiatic form, and spontaneous hypothyroidism had developed in this case.

Figure 4. Computed tomography scan showing bilateral extraocular (inferior reetus) muscle hypernophy.



Schwartz et al.[13] reported that the most prevalent form of dermopathy associated with GD was nonpitting edema (43.3%), and the elephantiatic form was found only 2.8% . In another review of 150 cases of dermopathy, the elephantiatic form occurred in only one patient [14]. Our patient had this rare form of dermopathy.

Presence of ophthalmopathy and dermopathy in Hashimoto's thyroiditis can be explained on the basis of sharing of antigens of thyroid (thyroid peroxidase, TPO) with orbital and dermal fibroblast. Circulating TSH-RAb activity was increased, while anti-TPO was elevated in this case. Wyse et al.[15] reported that TSH-RAb are also expressed in Hashimoto's thyroiditis. So, they suggested that the effect of TSH-RAb on orbital and dermal tissue remains uninhibited, resulting in development of orbitopathy and dermopathy. On the other hand, Cronin et al.[11] reported a similar case of Hashimoto's thyroiditis with opthalmopathy and pretibial myxedema, and they have also proposed that the TSH-RAb of GD which was raised in their case was ineffective in producing hyperthyroidism because of thyroid destruction due to Hashimoto's thyroiditis. Alternatively, hypothyroidism could have resulted from the effects of a circulating TSH-R blocking antibody. This case may be a Graves'disease which spontaneously culminated in Hashimoto's thyroiditis and hypothyroidism, but his medical history revealed that he had no any symptoms of hyperthyroidism. So, the association of elevated TSH-RAb levels and hypothyroidism can be explained as reported by Cronin et al.[11]. Yet it will be more correct to name it as severe dermopathy and orbitopathy wit autoimmune thyroid disease. Because the histopathology of the thyroid is not available to make a precise distinction between GD and Hashimoto's thyroiditis.

On the otherhand, there was no concomitant occurrence of Hashimoto's hypothyroidism and Thalessemia, as seen in our patient. Thus, this combination may be completely coincidental, since both disease are not rare in our country.

As a result, we here report a very severe dermopathy and ophthalmopathy associated with autoimmune thyroid disease.

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