Case Report

Medical Principles and Practice

Med Princ Pract 2013;22:408–410
DOI: 10.1159/000345212

Received: May 10, 2012
Accepted: October 15, 2012
Published online: November 22, 2012

A Case of Severe and Recurrent Painless Thyroiditis Requiring Thyroidectomy

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Key Words
Thyroid disease · Thyroid function · Thyroid surgery · Thyrotoxicosis

Abstract
Objective: To report a case of severe and recurrent painless thyroiditis requiring thyroidectomy. ***Clinical Presentation and Intervention:*** A 47-year-old man who presented with severe thyrotoxicosis was found to have extremely low radioactive iodine uptake, negative TSH receptor antibodies, and normal C-reactive protein; these findings suggested a diagnosis of painless thyroiditis. Due to the severity and recurrence of thyrotoxicosis, surgical resection of the thyroid gland was performed to prevent a thyrotoxic storm. Histological examination revealed typical lymphoid infiltration of the thyroid gland. ***Conclusion:*** This case illustrates that a patient with painless thyroiditis was successfully treated with surgery.
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Introduction

Painless thyroiditis is characterized by thyrotoxicosis, usually of mild to moderate severity, modest goiter, low radioactive iodine uptake, and negative thyroid-stimulating hormone (TSH) receptor antibodies. Histologically, the thyroid shows lymphocytic infiltration, at times organized into germinal centers, focal Hürthle cell changes, and/or focal hyperplasia in the thyroid epithelium surrounding the lymphocytes [1]. Painless thyroiditis can be confused with iodine-induced thyrotoxicosis (these patients, however, have pre-existing thyroid diseases such as multinodular goiter), or with factitious thyrotoxicosis (in this case, serum thyroglobulin is very low). The course of painless thyroiditis is usually transient with a hyperthyroid phase that lasts about 2 months before recovery, so that no treatment is typically required. The case presented here is unusual because of the recurrence and severity of the thyrotoxicosis, which required surgical resection of the thyroid gland to prevent a thyrotoxic storm.

Case Report

A 47-year-old man visited the outpatient clinic of Shinshu University Hospital because of recurrent thyrotoxicosis. Two weeks before the first admission, he consulted his physician because of palpitation and diarrhea. His clinical manifestations and thyroid function indicated profound thyrotoxicosis. Although TSH receptor antibody (TRAb) was negative, thiamazole was initiated because of severe thyrotoxicosis and related manifestations, including atrial fibrillation. The patient's thyroid function

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1011–7571/13/0224–0408$38.00/0

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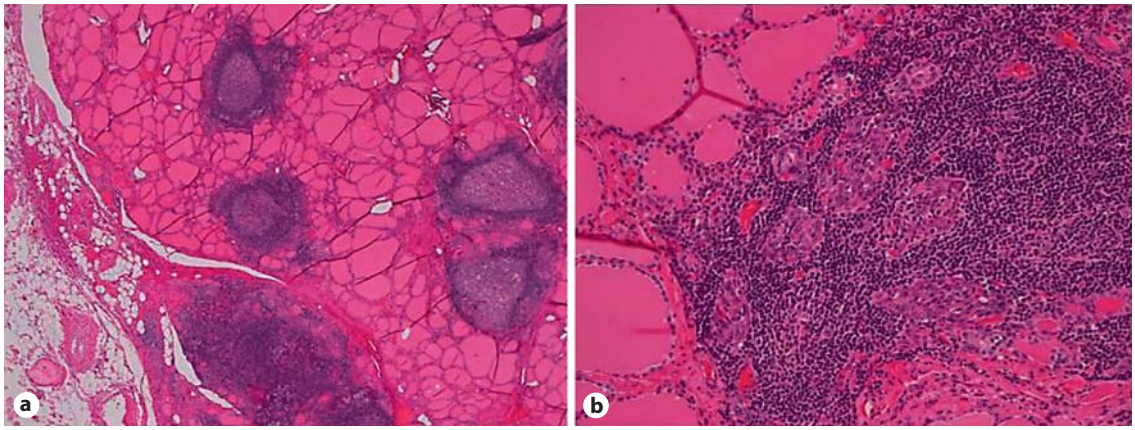

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Fig. 1. Pathology of the resected thyroid gland. HE staining showing lymphoid follicular formation with germinal center in the interstitium and lymphocyte infiltration in the follicular epithelium (**a**). Surrounding lymphoid follicular formation; lymphocyte infiltration was evident in the follicular epithelium (**b**).

and complaints improved within 3 weeks of commencement of thiamazole administration. After discontinuation of the antithyroid drug, hyperthyroidism recurred, and this cycle was repeated 3 times before the patient was referred to Shinshu University. His body weight decreased from 86 to 68.6 kg during the 8 months before admission. His physical findings on admission included low-grade fever (37–38°C), tachycardia (140–160 beats/min) with irregular rhythm, finger tremor, and excessive sweating. He suffered from severe diarrhea 3 weeks before the second admission. His thyroid gland was not enlarged and showed no spontaneous pain or tenderness. Complete blood count was unremarkable and C-reactive protein level was 0.66 mg/dl. The erythrocyte sedimentation rate was normal. The results of the endocrinological tests were as follows: TSH: 0.012 μIU/ml [normal value (NV): 0.20–4.00]; free T4: >7.77 ng/dl (NV: 1.00–2.00); free T3: >32.55 pg/ml (NV: 2.30–4.00); thyroglobulin: 8.4 ng/ml (NV: 0.0–78.0); anti-thyroglobulin antibody: 30.6 IU/ml (NV: 0.0–10.0); anti-thyroid peroxidase (TPO) antibody: 86.5 IU/ml (NV: 0.0–10.0); TRAb: 1.01 IU/l (NV: 0.00–1.30), and thyroid-stimulating antibody (TSAb): 91% (NV: <180%). Electrocardiography revealed atrial fibrillation and tachycardia. Chest X-ray showed no remarkable abnormalities. One week after the first admission, thyroid uptake was 0.38% (NV 4–16%), and the results of thyroid function tests were as follows: TSH: 0.007 μIU/ml; free T4: 6.76 ng/dl, and free T3: 8.83 pg/ml. Ultrasound showed no nodules in the thyroid gland, and blood flow was not remarkable. Total thyroidectomy was chosen as an appropriate treatment in this case to control severe thyrotoxicosis and prevent a thyrotoxic storm. Levothyroxine supplementation following resection controlled thyroid function. No relapse occurred after treatment. Diarrhea was improved. The pathological findings demonstrated lymphoid follicular formation with germinal center in the interstitium and lymphocyte infiltration in the follicular epithelium (fig. 1).

Discussion

The distinctive features of this case of painless thyroiditis were the recurrence of the thyrotoxicosis (7 times in 1 year) and its severity, which approached that of a thyrotoxic storm given the severe tachycardia and diarrhea. However, Hiraiwa et al. [2] reported a case of adrenal crisis with transient thyroiditis, in which another hormonal deficiency was not apparent. The diagnosis of painless thyroiditis was based on the presence of thyrotoxicosis with low radioactive iodine uptake and negative TSH receptor antibodies. Thyrotoxicosis factitia was included in the differential diagnosis but considered unlikely because the patient asserted that he never took thyroxine, thyroid gland extracts, or foods containing thyroid hormones. Even if the measurement of serum thyroglobulin was low, the histological appearance of the thyroid confirmed the autoimmune pathogenesis of thyrotoxicosis. The treatment of painless thyroiditis is typically limited to observation, given the transient and mild nature of the thyroid dysfunction. β-Adrenergic blockade is effective for the treatment of symptoms related to thyrotoxicosis. Antithyroid medication and iodide intake are not effective in preventing hormone release from the affected gland. At the beginning of management of this case, we treated with anti-thyroid drugs for a few weeks due to an initial diagnosis of Graves' disease, although TRAb was negative. Prednisolone is an alternative choice to treat painless thyroiditis, although steroid therapy was not initiated because of manic excitement and depressive states in this case. Drastic ablative therapy, such as surgery or radioac-

tive iodine, is rarely indicated. Although a sufficient absorbed radiation dose is required, radioactive iodide therapy was reported to be effective in some cases of recurrent painless thyroiditis [3, 4]. There have been three reports of painless thyroiditis treated by thyroidectomy [5, 6]. Although thyroidectomy is a unique therapy to achieve complete cure, physicians generally hesitate to recommend surgical resection for several reasons. First, this is a rare clinical course among common cases of painless thyroiditis. The second reason is that because thyrotoxicosis due to painless thyroiditis is a process of hypothyroidism, we predicted that thyrotoxic severity would subside. Third, exclusion of exogenous thyroid hormone intake is difficult. However, as shown in this case, surgical resection is the most effective form of treatment in cases of severe thyrotoxicosis.

Conclusion

This was a rare case of severe recurrent painless thyroiditis effectively managed by proactive surgical resection to avoid thyrotoxic crisis.

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