Case Report

Thyrotropinoma with Graves' disease detected by the fusion of indium-111 octreotide scintigraphy and pituitary magnetic resonance imaging

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ABSTRACT

Thyroid-stimulating hormone-secreting pituitary adenoma (TSHoma) is a rare benign endocrinological tumor which produces TSH in the pituitary gland. Herein, we presented a female patient having TSHoma with Graves' disease during and just after pregnancy that we found by indium-111 octreotide scintigraphy while investigating the patient for hyperthyroidism symptoms.

Keywords: Graves' disease, indium-111 octreotide scintigraphy, thyroid-stimulating hormone-secreting pituitary adenoma

INTRODUCTION

Thyroid-stimulating hormone (TSH)-secreting pituitary adenoma (TSHoma), also called thyrotropinoma, comprises 2% of all pituitary adenomas.[1] These tumors provoke central hyperthyroidism through releasing inappropriate TSH secretion.[2] TSHoma displays itself by hyperthyroidism symptoms most usually milder than the ones stemming from the thyroid or mass effects onto its neighbors.[3] Patients with this type of thyrotoxicosis can exhibit thyroid crisis because of stress or prior to surgery that must be intervened. Antithyroid drugs such as thiamazole afford to control hyperthyroidism relevant to TSHomas, but long-term use of these agents may induce expansion of the tumor over a negative feedback mechanism ending in a decrease of thyroid hormone levels.[1] The concomitant mixed pituitary adenoma secreting growth hormone and prolactin at the same time complicate the condition in the existence of a TSHoma accompanying thyroid disease.[4] Treatment modalities are surgery, radiation therapy, and somatostatin analogs. Somatostatin is a normally produced neuropeptide in the body. It acts over its receptors and these receptors are present in many tumors (mainly neuroendocrine tumors) and activated leukocytes of granulomatous and chronic inflammatory diseases.[5]

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CASE REPORT

A 37-year-old female was examined with thyroid function tests because of vague exophthalmos at the 20th week of her pregnancy. Free-T3 was found 8.96 pg/ml (normal range: 2.3–4.2 pg/ml), free-T4 2.79 ng/dl (normal range: 0.89–1.76 ng/dl), TSH 5.54 microIU/ml (normal range: 0.35–5.5 microIU/ml), and alpha-subunit of TSH 32 mIU/ml. Because of this inappropriate TSH secretion, a contrast magnetic resonance (MR) imaging was requested for a possible TSHoma. A macroadenoma of 13 mm × 11 mm in size adjacent to optic chiasm was detected in pituitary gland on MR. Her visual field was normal. She was treated by 3 × 50 mg/day of propylthiouracil orally and followed, for she did not accept the operation. She bore a healthy baby of full term and 3000 g in weight without any complication of this pituitary macroadenoma suspected of secreting TSH during her pregnancy and birth. Thyrotoxicosis and TSH level increased at postpartum 3rd month. These findings were

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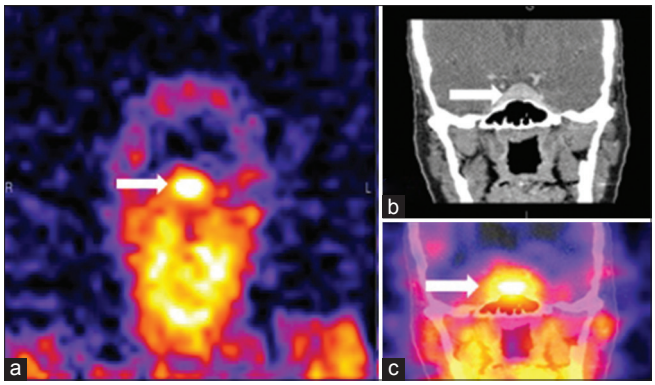

Figure 1: Indium-111 octreotide single photon emission computed tomography shows focal uptake in sella turcica consistent with thyrotropinoma on coronal single photon emission computed tomography (a), coronal magnetic resonance (b), and fused images of indium-111 octreotide and magnetic resonance (c)

strongly implying that macroadenoma in pituitary gland was a TSH-secreting tumor causing the hyperthyroidism symptoms of the patient. The patient stopped breastfeeding. A solid thyroid nodule of 11 mm × 10 mm in dimension, hypoactive on Tc-99m pertechnetate thyroid scintigraphy, was detected by ultrasonography and her TSH receptor antibody was 30 U/L (normal range <2 U/L). Fine-needle aspiration biopsy of the nodule revealed oxyphilic changes (Bethesda 4). Indium-111 (In-111) octreotide scintigraphy was performed to confirm the functional status of pituitary adenoma. Focal uptake within sella turcica consistent with the pituitary adenoma on MR [Figure 1] was seen and no other pathologic finding was observed on whole-body screening. She was operated with total thyroidectomy owing to malignancy suspicion. Surgical specimen was confirmed as oxyphilic nodular goiter and Graves' disease pathologically [Figure 2a]. TSH level increased after thyroidectomy. After that, she underwent transsphenoidal microscopic hypophysectomy later on. Immunohistochemical pathology reported the diagnosis as a TSH-secreting adenoma [Figure 2b].

DISCUSSION

Octreotide is a somatostatin analog which can be easily labeled with In-111 and is primarily used to detect neuroendocrine tumors.[6] It is used not only to diagnose these tumors but also to treat them. Somatostatin receptors are found in pituitary adenomas too.[7] Coexistence of TSHoma with Graves' disease is very rare and only six cases of such association with histological confirmation, all of whom are female, have been reported previously.[8-10] Our patient had signs of thyrotoxicosis, goiter, and exophthalmos with elevated thyroid hormone and TSH receptor antibodies, but normal TSH values. She was exhibiting all these typical diagnostic criteria and also had a mass in pituitary gland on MR strongly suggesting a thyrotropinoma. It was necessary to establish the functional status of this mass carrying strong evidence of TSHoma. However, her pregnancy precluded all invasive imaging modality and surgical interventions. It is possible to show TSHoma by In-111 octreotide scintigraphy if it is

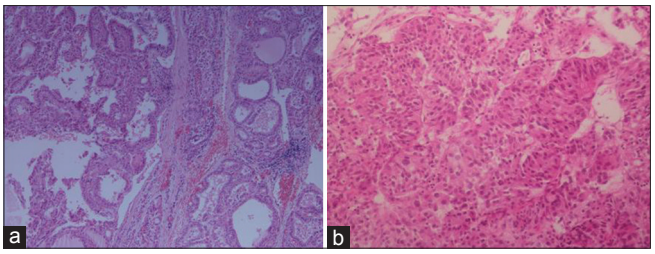

Figure 2: Graves' disease and oxyphilic nodular goiter (H and E, ×20) (a) and pituitary thyrotroph adenoma (H and E, ×40) (b)

somatostatin receptor positive. As hyperthyroidism symptoms and abnormality of biochemical markers persisted at postpartum period, we decided to show the functional relation between the abnormal laboratory tests causing patient's complaints and the mass by In-111 octreotide scintigraphy. We used fusion of octreoscan and MR for this purpose. Proving of this mass as a functional tumor with a focal uptake within sella turcica led to an ultimate decision of surgery reinforcing the hand of physician. A different and interesting feature of our case is the concomitant existence of Graves' disease and thyrotropinoma, which is different than the previous 6 cases of thyrotropinoma in the literature. In routine management of such a patient, pituitary surgery for macroadenoma is performed first, total thyroidectomy is performed later. However, in this case, total thyroidectomy was performed first because of malignancy suspicion.

CONCLUSION

Pituitary lesions causing inappropriate TSH secretion should be confirmed by functional imaging modalities. In-111 octreotide scintigraphy may be useful in illumination of such masses in pituitary gland, particularly in association of TSHoma with Graves' disease especially when TSH values are not compatible with either the clinical history or other thyroid functions tests.

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Conflicts of interest
There are no conflicts of interest.

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