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Case report

Hyperthyroidism due to struma ovarii: Diagnostic pitfalls and preventing thyroid storm



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ABSTRACT

We report struma ovarii in a case that had hyperthyroidism and was treated with laparoscopic tumor resection. A 40-year-old Japanese woman presented with tachycardia, finger tremor, and weight loss. Although blood examination showed hyperthyroidism, test results for thyroid stimulating hormone receptor antibody and thyroid stimulating antibody were negative, and thyroid scintigraphy showed no abnormal findings. Because she was diagnosed with an ovarian tumor, and whole-body scintigraphy showed that iodine uptake was detected in the pelvic space, we diagnosed her with an ovarian tumor, which caused excessive thyroid hormone secretion. After controlling the thyroid hormone level, we resected the ovarian tumor laparoscopically. The thyroid hormone level was within the normal range postoperatively without any medications. Based on our experience, physicians need to remember that ovarian tumors can cause hyperthyroidism. Controlling the thyroid hormone level preoperatively by using antithyroid drugs and performing minimally invasive laparoscopic surgery is considered useful for preventing thyroid storm.

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Introduction

Struma ovarii is an ovarian tumor defined by the presence of thyroid tissue comprising >50% of the overall mass. It most commonly occurs as part of a teratoma.^{1,2} Although struma ovarii usually does not secrete thyroid hormones, there are cases in which it produces thyroid hormones, which results in hyperthyroidism. When hyperthyroidism is caused by an ectopic thyroid hormone-producing tumor, it may be difficult to make a diagnosis, and few cases with hyperthyroidism due to struma ovarii have been reported. Here we report a case with hyperthyroidism due to struma ovarii that was diagnosed by whole-body scintigraphy, and treated with antithyroid drugs and laparoscopic tumor resection.

Case Report

A 40-year-old Japanese woman, gravida 2, para 0, had an unremarkable history. She visited a clinic of internal medicine because of tachycardia, finger tremor, and sweating, and she had a weight loss of 18 kg in the past 6 months. Although blood examination showed hyperthyroidism (free triiodothyronine, 10.4 pg/mL; free thyroxine, 3.8 ng/dL; and thyroid stimulating hormone, < 0.01 μIU/mL), both the thyroid stimulating hormone receptor antibody and thyroid stimulating antibody were negative. Furthermore, thyroid ultrasonography showed no abnormal findings, and ^{99m}Tc thyroid scintigraphy showed no abnormal uptake to the thyroid (Figure 1A). Although she was advised to take antithyroid drugs before a diagnosis was made, she refused and stopped visiting the clinic.

Two months later, she was diagnosed with a left ovarian polycystic tumor measuring 7 cm in diameter by cancer screening, and she was referred to our hospital. Further examination of antithyroid antibodies showed that the antithyroglobulin antibody was positive (71.3 IU/mL) and antithyroid peroxidase antibody was negative. The tumor was suspected to be an ovarian mature cystic teratoma

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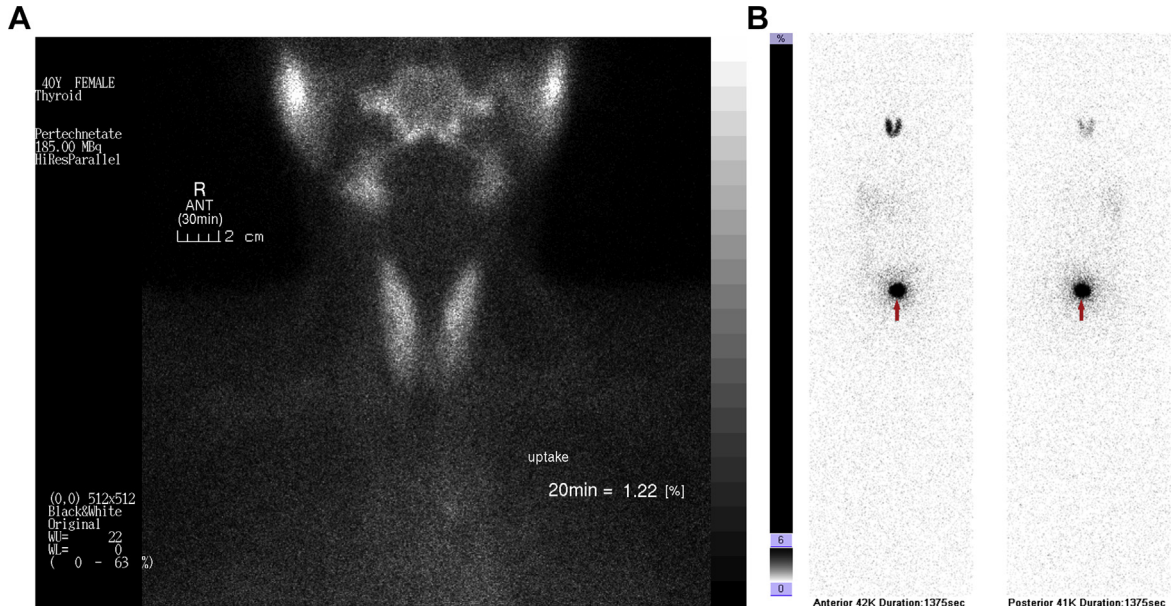


Figure 1. (A) ^{99m}Tc thyroid scintigraphy shows no abnormal uptake to the thyroid; (B) ^{123}I whole-body scintigraphy shows a low radioactive iodine uptake to the thyroid gland and a high iodine level in the left pelvic space.

by magnetic resonance imaging, and it was suspected to be functional for producing thyroid hormone. ^{123}I whole-body scintigraphy showed a low radioactive iodine uptake to the thyroid gland, and a high iodine level was present in the left pelvic space (Figure 1B).

From these observations, she was diagnosed with an ovarian tumor, which caused excessive thyroid hormone secretion. We decided to operate after controlling the thyroid hormone level to prevent thyroid storm, which can be caused by surgical invasion. Treatment was started with oral methimazole (MMI; 15 mg/d) and potassium iodide (50 mg/d). A rash on the skin caused by MMI developed, so we replaced the existing treatment with propylthiouracil (PTU; 200 mg/d) and potassium iodide (50 mg/d) 2 weeks later. Because thyroid hormone level was controlled for 6 weeks, she underwent laparoscopic left salpingo-oophorectomy at 7 weeks after the initiation of medical treatment.

The thyroid hormone level was maintained within the normal range postoperatively without any medications, and she was discharged from the hospital 3 days postoperatively without any

complications. Histological examination showed that the tumor was a mature cystic teratoma with a thyroid tissue component exceeding 50%. Therefore, we diagnosed it as struma ovarii. After surgical resection of the struma ovarii, the patient became euthyroid (Figure 2).

Discussion

The main conclusions from this case are as follows: struma ovarii may be functional; ^{123}I whole-body scintigraphy is useful for diagnosis; prior to surgery, controlling the thyroid hormone level is necessary to prevent thyroid storm, which may be caused by surgical invasion; and laparoscopic surgery may be useful for preventing thyroid storm so minimally invasive surgery can be performed.

Although it is uncommon for struma ovarii to produce thyroid hormone, which results in hyperthyroidism,^{2,3} there are some reported cases.^{4,5} For diagnosis, it is important to suspect a functional

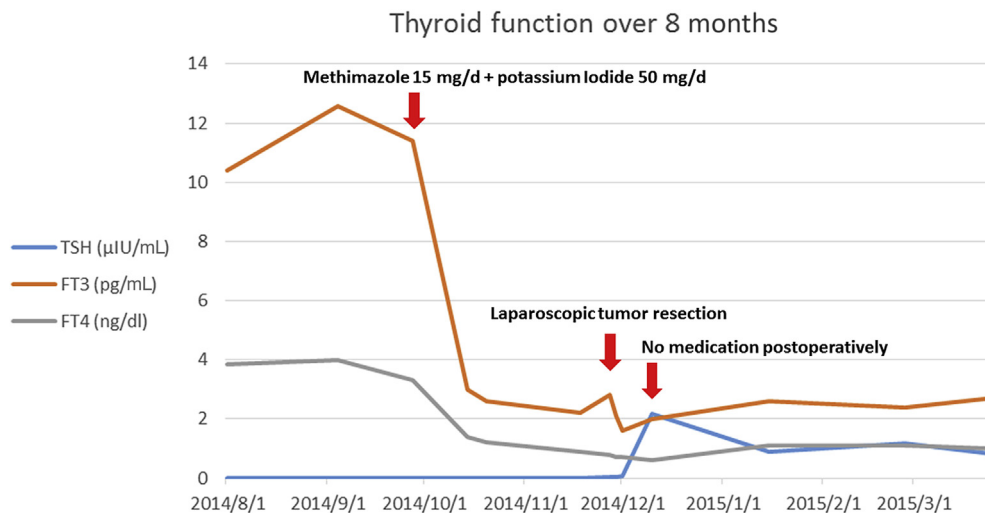


Figure 2. Thyroid function over 8 months. FT3 = free triiodothyronine; FT4 = free thyroxine; TSH = thyroid stimulating hormone.

ovarian tumor in a patient with hyperthyroidism. As thyroid scintigraphy is insufficient to make a diagnosis, physicians need to perform whole-body scintigraphy. When patients have a thyroid disease due to Graves' disease and a unilateral functioning struma ovarii concurrently, caution should be exercised.⁴ Additionally, physicians must be careful about diagnosing a functional struma ovarii because a case of hyperthyroidism due to a malignant struma ovarii has been reported.⁵

Both MMI and PTU are effective for controlling the thyroid hormone level, and surgically removing the tumor normalizes this level in patients with a functional struma ovarii. Antithyroid drugs must be properly given because the most common trigger of thyroid storm has been reported to be the irregular use or discontinuation of antithyroid drugs.⁶ In our case, antithyroid drugs were formulated until the day before surgery, and they were discontinued from the date of surgery. Because the thyroid hormone level immediately returns to within the normal range after surgery, physicians need to stop antithyroid drugs and monitor the patient for hypothyroidism postoperatively.^{2,7,8} These drugs should not be carelessly prescribed postoperatively.

Unexpected emergency surgery for torsion or rupture of the tumor should be avoided, and elective laparoscopic surgery is recommended because invasive surgery may cause thyroid storm. In general, laparoscopic surgery has increased significantly for benign ovarian tumors in recent years,⁹ and it is the gold standard for benign ovarian tumors because of its overall improved quality of life.¹⁰ Therefore, laparoscopic surgery should be considered for the treatment of benign ovarian tumors. To prevent thyroid storm during surgery, a shorter operation time is recommended, and pain must be completely controlled. In the current case, the tumor was retrieved in a bag because leakage due to rupture of the tumor may have resulted in an increased blood level of the thyroid hormone. Additionally, to prevent thyroid storm postoperatively, a transversus abdominis plane block was used to suppress pain. As nonsteroidal anti-inflammatory drugs induce an increase in the serum thyroid hormone level,¹¹ we used acetaminophen for pain relief postoperatively.

Mature cystic teratoma is a common ovarian tumor; therefore, some ectopic thyroid hormone-producing tumors may remain

unrecognized, or they are operated on as nonfunctioning tumors. Physicians must be careful to diagnose mature cystic teratoma because it is sometimes associated with other rare tumors such as peripheral primitive neuroectodermal tumor.¹² Therefore, diagnosis of ovarian mature cystic teratoma may have several possible diagnostic pitfalls. Physicians need to exercise caution, especially when emergency surgery is required for patients with a mature cystic teratoma, because it may secrete thyroid hormone.

References

1. Wei S, Baloch ZW, LiVolsi VA. Pathology of struma ovarii: a report of 96 cases. *Endocr Pathol*. 2015;26:342–348.
2. Laganà AS, Santoro G, Triolo O, Giacobbe V, Certo R, Palmara V. Hashimoto thyroiditis onset after laparoscopic removal of struma ovarii: an overview to unravel a rare and intriguing finding. *Clin Exp Obstet Gynecol*. 2015;42:673–678.
3. Wang Y, Pan L, Huang H, Shen K, Wu M, Lang J. Clinical study on 68 cases with struma ovarii. *Zhonghua Fu Chan Ke Za Zhi*. 2014;49:451–454 [In Chinese, English abstract].
4. Sitasuwan T, Hanamornroongruang S, Peerapatdit T, Thongtang N. Coexistence of Graves' disease and unilateral functioning struma ovarii: a case report. *BMC Endocr Disord*. 2015;15:68.
5. Matsuda K, Maehama T, Kanazawa K. Malignant struma ovarii with thyrotoxicosis. *Gynecol Oncol*. 2001;82:575–577.
6. Akamizu T, Satoh T, Isozaki O, et al. Diagnostic criteria, clinical features, and incidence of thyroid storm based on nationwide surveys. *Thyroid*. 2012;22:661–679.
7. Carvalho JP, Carvalho FM, Lima de Oliveira FF, Asato de Camargo RY. Hypothyroidism following struma ovarii tumor resection: a case report. *Rev Hosp Clin Fac Med Sao Paulo*. 2002;57:112–114.
8. Amareen VN, Haddad FH, Al-Kaisi NS. Hypothyroidism due to Hashimoto thyroiditis post struma ovarii excision. *Saudi Med J*. 2004;25:948–950.
9. Wu MP, Lee CL. The trends of minimally invasive surgery for benign gynecologic lesions, 1997–2007 in Taiwan. *Gynecol Minim Invasive Ther*. 2012;1:3–8.
10. Kotani Y, Umemoto M, Tobiume T, Shiota M. Ovarian tumor cases that were preoperatively diagnosed as benign but postoperatively confirmed as borderline or malignant after laparoscopic surgery. *Gynecol Minim Invasive Ther*. 2013;2:122–125.
11. Lim CF, Bai Y, Topliss DJ, Barlow JW, Stockigt JR. Drug and fatty acid effects on serum thyroid hormone binding. *J Clin Endocrinol Metab*. 1988;67:682–688.
12. Huang BS, Horng HC, Lai CR, et al. Peripheral primitive neuroectodermal tumor of the ovary with torsion. *Gynecol Minim Invasive Ther*. 2013;2:65–69.