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Struma Ovarii co-incident with Hashimoto's thyroiditis: an unusual cause of hyperthyroidism

Kelly Morrissey^a, Craig Winkel, MD^b, Sheri Hild, PhD^c, Ahalya Premkumar, MD^d, and Pamela Stratton, MD^a

^aReproductive Biology and Medicine Branch, National Institute of Child Health and Human Development, Bethesda, Maryland ^bDepartment of Obstetrics and Gynecology, Georgetown University Medical Center, Washington, DC ^cBioqual, Rockville, Maryland ^dDepartment of Radiology, Clinical Center, National Institutes of Health, Bethesda, Maryland

Abstract

Case summary—Struma ovarii is a rare type of ovarian teratoma comprised of mainly of thyroid tissue that does not typically alter thyroid hormone levels and uncommonly recurs or persists after ovarian cystectomy. We report a case of struma ovarii causing hyperthyroidism, co-incident with Hashimoto's thyroiditis. The patient had a history of asymptomatic struma ovarii found at laparoscopic surgery, which had been removed by ovarian cystectomy 3 years earlier. In a patient with two different causes of abnormal thyroid function, it is important to seek an encompassing clinical scenario; in this instance, we identified an extraglandular cause of hyperthyroidism concomitant with an underactive thyroid gland.

Objective—To report the identification of struma ovarii in a patient with a history of struma ovarii and new hyperthyroidism

Design—Case report

Setting—Academic research hospital

Patient(s)—A woman with hyperthyroidism who has struma ovarii, co-incident with Hashimoto's thyroiditis

Interventions—Laparoscopic salpingo-oophorectomy

Main Outcome Measure(s)—Measurement of thyroid hormone parameters before and after surgery

Result(s)—After removal of the second struma ovarii, hyperthyroidism resolved.

Conclusion(s)—In a patient with two different causes of abnormal thyroid function, it is important to seek an encompassing clinical scenario

Reprint Requests: Pamela Stratton, MD, National Institutes of Health, 10 Center Dr. MSC 1109, Bldg 10-CRC Rm 1-3140 Bethesda, MD 20892-1109, (Fax: 301 480-6703; email: ps79c@nih.gov).

We describe a patient who developed new clinical hyperthyroidism coincident with Hashimoto's thyroiditis; a history of struma ovarii facilitated the diagnosis of an ovarian tumor, and the ovary was removed.

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Keywords

struma ovarii; ovarian neoplasms; hyperthyroidism

Introduction

Struma ovarii is an uncommon type of ovarian teratoma composed predominantly of mature thyroid tissue which results in hyperthyroidism in less than 10% of cases (1-3). Approximately 95% of strumas are benign, and preoperative clinical or radiological diagnosis of struma ovarii is very difficult because they can be small and asymptomatic (1,2). We describe an incidental finding of struma ovarii first recognized on MRI as a complex mass, removed at laparoscopy for endometriosis. Three years later, a struma ovarii was found again in the same ovary and was accompanied by hyperthyroidism. The prior history of struma ovarii helped guide the diagnosis of ovarian tumor when the clinical symptoms of hyperthyroidism occurred.

Case Report

A 38-year-old woman with previous history of surgically diagnosed endometriosis presented with a 7-year history of worsening pelvic pain to participate in an IRB-approved study of the treatment of endometriosis and chronic pelvic pain. On review of systems at the first study visit, the patient related a history of intermittently elevated levels of serum thyroxine. Evaluation by an endocrinologist, however, indicated no thyroid abnormalities. At study entry, both serum thyroxine and thyroid stimulating hormone levels were normal.

The double-masked, placebo-controlled clinical study of chronic pain associated with endometriosis included laparoscopic surgical excision of endometriosis followed by treatment with a raloxifene or placebo for six months, with a second laparoscopy when chronic pelvic pain returned, or at two years. Prior to the first surgery, a pelvic MRI was performed as a research study to evaluate the utility of MRI in diagnosing endometriosis; a mid-luteal phase ultrasound was performed for comparison later on hormonal treatment. The ultrasound revealed two 2-3 cm cysts in the right ovary. One had a perivascular pattern consistent with ovulation; the other was homogeneous without vascularization and presumed to be an endometrioma. MRI showed a 3 cm mass adjacent to a right ovarian cyst (Figure 1 and 2).

At laparoscopy to remove endometriosis, the ovary was mobile and smooth but asymmetric with a firm mass at one pole. The mass was excised using a contact tip laser (Surgical Laser Technologies, The Oaks, Pennsylvania) and was believed to be benign based on its smooth mobile appearance. Bleeding was easily controlled with bipolar cautery forceps. Histological examination was consistent with benign struma ovarii. Endometriosis was also excised and the patient was randomized to placebo.

At the time of study laparoscopy two years later to again remove endometriosis, the right ovary was mobile and appeared to be normal; thyroid function tests were also normal.

A year later, the patient had symptoms consistent with hyperthyroidism and, on examination, had a slightly enlarged thyroid. During evaluation, the internist placed her on propranolol. Serum T4 levels were elevated (16.8 ug/ml, normal range 4.5 to 12.5), TSH (< 0.40, normal range 0.40-4.00) was completely suppressed, and antibodies to thyroglobulin and thyroid peroxidase were positive.

Thyroid uptake and imaging, and a pelvic ultrasound were done. The thyroid scan was normal and the iodine uptake was very low, only 2%. A small thyroid nodule (0.5 cm) was subsequently biopsied and histopathology was consistent with Hashimoto's thyroiditis. Pelvic ultrasound

showed a 3 by 4 cm complex cystic mass in the right ovary, with an appearance consistent with a dermoid cyst. On transvaginal ultrasound and pelvic examination, the right ovarian mass persisted over three to four months. The CA-125 level was normal.

She continued to have symptoms until four months later when the T4 levels were noted to be in the high normal range. At laparoscopy, an ovarian tumor was again noted and a right salpingo-oophorectomy was performed. Peritoneal washings showed no malignant cells. On histological evaluation, the ovarian tissue was exclusively differentiated into follicular thyroid tissue and described as a benign struma ovarii (figure 3). Hyperthyroidism resolved after removal of the ovary.

Discussion

We describe struma ovarii coincident with symptomatic hyperthyroidism which occurred three years after we removed a non-functioning struma ovarii during laparoscopic surgery for endometriosis. This rare type of teratoma is comprised primarily of thyroid tissue but does not result usually in altered thyroid hormone levels(2). While in one series, five of 25 patients had evidence of hyperthyroidism (3), a review by Smith (4) placed the rate much lower, with only 17 of 152 having thyrotoxicosis. As others have noted, the preoperative clinical or radiologic diagnosis of struma ovarii is very difficult except when the tumor is very large or there are associated changes in thyroid function (1,3,4). For this patient, because of the history of struma ovarii, the early and accurate diagnosis was made by performing an ultrasound.

It is exceedingly uncommon to have two simultaneous causes of abnormal thyroid function and illustrates the importance of seeking a unifying clinical scenario to explain the laboratory and radiographic findings. Others have inadvertently removed the thyroid before the ovarian tumor was found (3). In this case, despite a recent elevation of serum T4 levels and symptoms of thyrotoxicosis, laboratory and radiographic tests revealed low thyroid iodine uptake and laboratory markers of Hashimoto's thyroiditis. It is possible that the struma ovarii resulted in the formation of anti-thyroid antibodies and thus result in secondary thyroiditis.

Though destructive autoimmune thyroiditis can result in hyperthyroidism that spontaneously resolves, the decrease in T4 immediately following removal of the tumor combined with the histologic findings of a struma ovarii suggests that the ovarian tumor was an extraglandular cause of hyperthyroidism. A radioiodine scan of the pelvis might have been useful in diagnosing a struma ovarii producing significant amounts of thyroid hormone (5), but was not done.

The radiographic findings on ultrasound and MR imaging, before the first surgery, of a mass adjacent to an ovarian cyst were attributed to endometriosis. Since endometriomas can appear as masses, the clinical suspicion of an ovarian tumor was low. When the tumor was noted at surgery, the differential diagnosis included benign solid ovarian tumors such as teratoma and fibrothecoma (6). On MR imaging, immature teratomas and solid mature teratomas share characteristic appearances that makes them indistinguishable radiologically (7). While small cystic spaces which show either low or high signal intensity on MRI are characteristic of struma ovarii, a cyst adjacent to a solid mass was noted on the preoperative MRI (1,7).

The most common ovarian teratoma, mature cystic teratoma, is similar to struma ovarii in that both are often unilateral and occur in women of all ages (6 to 74 years)(3,4). Unlike struma ovarii which is difficult to diagnose preoperatively, mature cystic teratoma may be diagnosed radiologically because teeth, bone and cartilage in addition to fatty material and hair are readily detectable with imaging techniques (8). However, it too is often discovered as an incidental finding on physical examination, radiologic examination, or at an unrelated abdominal operation (8).

Both struma ovarii tumors arose from the same ovary and appeared benign, both at surgery and on histology; it is unclear whether this is a case of recurrent or multiple struma ovarii. While the risk of recurrent struma ovarii is not known, the risk of recurrent dermoid was reported in 3 of 99 women in one series who underwent ovarian cystectomy (9). Since only the ovarian tumor was removed at the first laparoscopy, the tumor was likely persistent rather than recurrent. Even though the struma ovarii extended to the margin of the ovarian tissue removed at the first surgery, the remaining ovarian tissue appeared normal and additional surgery was not considered. A small nest of residual tumor was not clinically apparent at a second laparoscopy performed when the patient was asymptomatic and had normal thyroid levels.

The removal of an ovarian mass at surgery performed for endometriosis illustrates the importance of systematic inspection of the pelvis and reproductive organs at operative laparoscopy. Since we routinely obtain consent to remove unexpected, abnormal findings that do not compromise the patient's reproductive function, we were able to successfully diagnose this uncommon ovarian tumor. The persistence of struma ovarii is also uncommon, but might have been suspected because of struma ovarii at the histologic margin. Most importantly, however, because of the history of struma ovarii, when the patient developed new clinical hyperthyroidism in the presence of Hashimoto's thyroiditis, the diagnosis of ovarian tumor was made and the ovary was removed.

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Figure 1.

Figure 1A: Sagittal T2 weighted MRI of the pelvis showing a low signal intensity solid mass (arrow) adjacent to a cyst in the right ovary.

Figure 1B: Axial T2 weighted MRI of the pelvis showing a low signal intensity solid mass (arrow) and a cyst in the right ovary.

Figure 1C: Hematoxylin and eosin stained photomicrograph of right ovary with struma ovarii at 40 times magnification with calibration bar.