Struma Ovarii: Case Report and Review

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Abstract. Only 150 cases of struma ovarii (i.e., goiter of the ovary) have been reported in the medical literature. The condition is characterized by the presence of thyroid tissue in an ovarian tumor, typically as a part of a teratoma. We report the first case of Hashimoto's thyroiditis concurrent with struma ovarii, a thyroid gland with Hürthle cells, and positive antithyroid antibodies despite reference-range thyroid function test results. **Case Report**. A young woman presented to the office with pelvic pain. Ultrasound revealed a complex right ovarian mass. Her TSH levels was elevated. The mass was surgically removed, and biopsy revealed struma ovarii with Hashimoto's thyroiditis. After surgery, the patient's TSH level decreased into the reference range, and her free T_4 and free T_3 levels remained within their reference ranges. However, her thyroid peroxidase and antithyroglobulin levels are elevated. She continues to remain asymptomatic with reference-range thyroid function of struma ovarii in young patients who present with an ovarian mass, as resolution of thyroid function abnormalities can be a-chieved by surgical removal of the ovarian mass.

Keywords Hashimoto's thyroiditis • Hürthle cells • Immunohistochemical stain • Struma ovarii • Teratoma

Introduction

Struma ovarii is a rare entity, with only 150 reported cases in the medical literature. It is characterized by the presence of thyroid tissue in an ovarian tumor, typically as a part of a teratoma.^[2]

Struma ovarii was first described by Von Kalden in 1895 and Gottschalk in 1899.^[1] Most struma ovarii are benign with only 5%-to-10% being malignant.^[3] Hyperthyroidism is seen in about 8% of patients^[4] with struma ovarii, but most cases of struma ovarii are asymptomatic from a thyroid standpoint with the majority of patients presenting with symptoms attributable to mass effect, such as pelvic pain.

The pathophysiology has not been clearly defined. Struma ovarii can be identified on histological exam. In difficult cases, immunohistochemical stains for thyroid transcription factor and/or thyroglobulin will confirm the diagnosis of struma ovarii.^[5] The diagnosis is usually made after surgical resection of the pelvic tumor. Rarely struma can have characteristics of Hashimoto's thyroiditis. We hereby report a very rare case of struma ovarii presenting with abnormal thyroid function studies due to Hashimoto's thyroiditis.

CASE REPORT

A 48-year-old woman was seen by her gynecologist for mild pelvic pain of insidious onset. Her abdominal and pelvic examinations were noted to be unremarkable. However, pelvic ultrasound showed a complex right ovarian mass. She subsequently underwent a laparoscopic right salpingo-ophorectomy.

The pathology of the surgical specimen revealed a dermoid cyst with a 1 cm tan-white mural nodule. Histological examination of the nodule showed Hürthle cells with abundant pink cytoplasm, nuclear pleomorphism, and prominent nucleoli (Figure 1). The sheets of cells were infiltrated with lymphocytes. Foci of cells forming follicles were identified at the periphery of the nodule.

Immunohistochemical stains revealed the cells to be positive for thyroid transcription factor stain (Figure 2). These findings are consistent with Hashimoto's thyroiditis arising within an ovary, which is designated "struma ovarii." The patient was then referred to our clinic for endocrine evaluation.

Her medical history revealed that she was diagnosed with thyromegaly after the birth of her first child approximately 18 years before. She had been on thyroid hormone therapy for two years until the birth of her second child. At her initial visit, she denied symptoms of thyroid dysfunction and further denied any local neck symptoms referable to her thyroid. She was not aware of any anterior neck/thyroid enlargement at the time of the visit. However, physical examination her thyroid gland was performed and revealed that the gland was minimally enlarged, firm, and had a bosselated texture consistent with Hashimoto's thyroiditis. Preoperative laboratory testing revealed that her serum thyroid stimulating hormone level was 5.73 uIU/mL (0.35-to-5.5 uIU/mL) with referance-range T_4 and T_3 levels.

Immediately after the surgery, the patient's serum TSH was 3.33 uIU/mL (0.35-to-5.5 uIU/mL)



Figure 1. Follicles with Hürthle cell change and infiltrating lymphocytes. Low mag (10x).

and remained within the reference range at 4-months follow-up. Her antithyroglobulin antibody titer was 851 IU (0-to-24 IU) and her antithyroid peroxidase antibody titer was 1492 IU (0-to-4 IU). A thyroid ultrasound showed a diffusely heterogeneous sonographic appearance, which was interpreted as consistent with Hashimoto's thyroiditis. One year following her ovarian surgery, the patient remains asymptomatic and her serum TSH remains is within the reference range.

DISCUSSION

Struma ovarii with pathologic evidence of autoimmune thyroiditis is uncommon. In 1965, Erez et al.^[6] described the first case of chronic thyroiditis in a 41-year-old with benign cystic teratoma of the ovary. In their case, the patient had undergone a partial thyroidectomy for chronic thyroiditis. Hashimoto's disease was not specifically mentioned in the pathology report from the partial thyroidectomy. The

patient was later found to have bilateral benign cystic ovarian teratomas. In the ovarian thyroid tissue, chronic thyroiditis was found consistent with Hashimoto's disease. Postoperative tests of thyroid antibody activity were negative.

In 1972, Watson^[7] reported only one case of Hashimoto's disease of the thyroid component of a benign cystic ovarian teratoma after reviewing three



Figure 2. Photo labeled TTF1: Immunohistochemical stain for TTF-1 reveals positive nuclear staining, confirming thyroid origin.

hundred and fifteen cases of ovarian teratomas. In this cases, there was no prior thyroid surgery and the patient's thyroid exam was normal. Immunological investigations failed to demonstrate the presence of any thyroid-component antibodies one year after the operation.

In 1991, Farrell^[8] reported a case of Hashimoto's disease in a benign cystic teratoma of an ovary in a 14-year-old with positive thyroid specific antibodies. In 1998, Fernando et al.^[11] reported a case of histological autoimmune thyroiditis in struma ovarii, but Hürthle cells were not seen.

Carvalho in 2002^[9] and Amareen in 2004^[10] reported cases of Hashimoto's thyroiditis and frank hypothyroidism following the resection of struma ovarii tumors. We report the first case of Hashimoto's thyroiditis involving struma ovarii and a thyroid gland

with Hürthle cells, positive antithyroid antibodies, but normal thyroid function.

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