International Electronic Journal of Medicine

Int Electron J Med. 2019; 8(2):140-142

doi 10.34172/iejm.2019.12

Case Report

Struma Ovarii With the Coexistence of Hashimoto's Thyroiditis: A Case Report

Zahra Abbasi Ranjbar[1], Seyedeh Hajar Sharami[2*], Ali Moradi Nakhodchari[3], Seyedeh Fatemeh Dalil Heirati[2]

[1]Reproductive Health Research Center, Department of Endocrinology and Metabolism, Al-Zahra Hospital, School of Medicine, Guilan University of Medical Sciences, Rasht, Iran.
[2]Reproductive Health Research Center, Department of Obstetrics and Gynecology, Al-Zahra Hospital, School of Medicine, Guilan University of Medical Sciences, Rasht, Iran.
[3]Department of Pathology, Guilan University of Medical Sciences, Rasht, Iran.

Abstract

Background: Struma ovarii (SO) is a rare phenomenon. In addition, it can be identified from the histological assessment and may show the characteristics of Hashimoto's thyroiditis in rare cases. In several cases, malignant stroma ovarii is found to produce hyperthyroidism while very few cases are reported with Hashimoto's thyroiditis. The present study reported SO co-incident with Hashimoto's thyroiditis. It should be mentioned that accurate monitoring of any signs of hypothyroidism is highly important after the surgery.

Case Report: A 21-year-old patient with oligomenorrhea, hypogastric pain for the last year, dyspareunia and dysmenorrhea with a right adnexal cystic focus, and a bright echogenic focus referred to Al-Zahra hospital, Rasht. Histopathologic examination was compatible with the diagnosis of mature cystic teratoma containing SO co-incident with Hashimoto's thyroiditis while the patient was in a good general appearance 2 days after the surgery. Regarding the subclinical hypothyroidism and due to her willingness to conceive, the clinician administered levothyroxine and thus the patient was in a stable state within 6 months.

Conclusion: In general, it is recommended to consider SO in women with ovarian mass and impaired thyroid function.

Keywords: Struma ovarii, Hashimoto's thyroiditis, Case report

***Correspondence to**
Seyedeh Hajar Sharami, Reproductive Health Research Center, Department of Obstetrics and Gynecology, Al-Zahra Hospital, School of Medicine, Guilan University of Medical Sciences, Rasht, Iran
Tel: 013-33369224
Email: sharami@gums.ac.ir

Received: October 2, 2019, Accepted: December 8, 2019, ePublished: December 15, 2019

Introduction

Struma ovarii (SO) is considered as a rare phenomenon and a specialized or monodermal teratoma predominantly composed of mature thyroid tissue. (1) Thyroid tissue must comprise more than 50% of the overall tissue to be classified as a SO. In addition, SO accounts for approximately 5% of all ovarian teratomas (2, 3). Depending on the histologic features, SO can be classified as benign or malignant (4). Women with SO usually present with pain and/or a pelvic mass and less frequently with ascites. (4, 5) Further, the histologic features of thyroid cancer are found in 5 to 37% of SO (3, 6). Cancer is more likely in larger tumors and rarely in tumors under 5 cm (7). In women presenting with a pelvic mass, SO is typically diagnosed postoperatively based upon the histologic findings of thyroid follicles in the resected ovary. Oophorectomy is regarded as the initial treatment of SO and patients with malignant SO may need therapy in addition to oophorectomy, particularly in the presence of metastatic disease (4).

Although it is commonly composed of normal thyroid tissue, the concurrent lymphocytic infiltration of the thyroid gland, as found in Hashimoto's thyroiditis and the ectopic thyroid tissue are extremely rare cases (8, 9).

In several cases, malignant SO was reported to produce hyperthyroidism (10) although there is no reported case of acute hypothyroidism following SO tumor resection. Considering the above-mentioned explanations, the present study aimed to report a very rare case of SO with the coexistence of Hashimoto's thyroiditis.

Case Presentation

A 21-year-old patient with oligomenorrhea, hypogastric pain for the last year, as well as dyspareunia and dysmenorrhea, trying to conceive after 2 years of marriage referred to Al-Zahra hospital, Rasht. The patient had no infertility assessment and had been taking HD (High

dose combined oral contraceptive pill) for 2 months in the last 5 months in order to treat the ovarian cysts.

Although no medical record was noted, thyroid disorder was reported in her first-degree family. Moreover, there was no evidence of abnormality during hip physical examination.

During the ultrasound examination, a right adnexal cystic focus (39 * 54 * 74 mm) with a bright echogenic focus (39 * 46 * 48 mm) was observed and indicated as the dermoid cyst.

The patient was hospitalized for laparotomy-cystectomy and tumor markers were analyzed as well. The results showed Ca125 = 11.6, CEA = 1.1, and AFP = 1.2. During laparotomy in the right ovary, a 5 * 5 dermoid cyst was evacuated and a 2 * 2 simple cyst was cauterized and evacuated in the left ovary. Then, the patient was followed up and the samples were transferred to pathology. The histopathologic examination of the cyst demonstrated the tumoral proliferation of mature tissues. Normal derivatives of more than 1-germ layers, including cartilage, adipose, neural, and salivary glands were noted as well. Furthermore, there was a component of the thyroid tissue with marked lymphocytic infiltration, along with lymphoid follicle formations, reactive germinal centers, and some thyroid follicular cells with the Hürthle cell change. It was compatible with the diagnosis of mature cystic teratoma containing SO co-incident with Hashimoto's thyroiditis (Figures 1 and 2).

Two days after the surgery, the patient was in a good general appearance and thus she was discharged after administering oral cefixime and metronidazole. Then, she was referred to the endocrinology clinic for further assessment. Laboratory results showed 410 units per mL thyroid peroxidase antibody, 7.58 mm units per mL thyroid-stimulating hormone, 13.2 free T4, and 2.33 free T3. Regarding the subclinical hypothyroidism and due to her willingness to conceive, the clinician administered levothyroxine. Finally, thyroid ultrasound was normal and the patient was in a stable state within 6 months.

Discussion

SO is a monodermal ovarian teratoma that contains a large amount of thyroid tissue, which may show diverse pathologic and histologic patterns such as normal, adenoma, along with papillary and follicular carcinoma. The probability of the malignancy of SO is rare and about 5% may be malignant. Additionally, metastatic disease may occur in 5%-6% of cases (11). These tumors are often found by non-specific morphology and it is most difficult to detect unless the tumor is extremely large or there are obvious signs of thyrotoxicosis (1). In addition, they are commonly diagnosed by the pathologic sample after cystectomy or hysterectomy (12). In half of the patients, ovarian tumors may be detected accidentally on ultrasound. Although the diagnostic criteria for SO is based on the histologic appearance of the ovarian sample, it is sometimes necessary to determine the origin of the cell by immunohistochemistry staining for thyroglobulin (13). In the recent report which was similar to previous investigations (8, 9, 13-15), the probability of a dermoid cyst was noted in the sonographic report, which was finally diagnosed as SO.

Although SO can be detected at any age even in childhood, it is mostly observed in the reproductive age (16). In the recent report, the patient was in a reproductive age, which is similar to other previous evaluations (8, 9, 13-15). However, some reports of the ovarian stroma are available in the 5th and 6th years of life or under 20 years (8, 9, 15).

The size of the ovarian stroma may vary, but they are generally less than 10 cm and often one-sided. In this study, there was a 5 cm cyst in the right ovary. Previous reports also mentioned one-sided cysts smaller than 10 cm (9, 13, 15). Contrarily, larger sizes for cysts (20 and 30 cm) were also noted in other previous reports (11, 16, 17). In addition, mass size is commonly important in determining the risk of malignancy and cancer is more likely in larger tumors (75% of tumors greater than 16 cm) and rare in tumors under 5 cm (7).

In several cases, malignant stroma ovarii was shown to produce hyperthyroidism (10), but there is no reported case of acute hypothyroidism following stroma ovarii tumor resection.

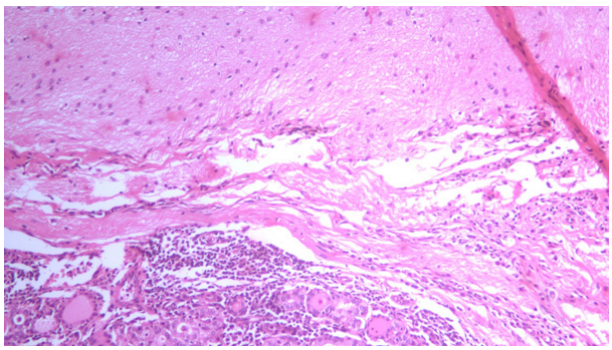

Figure 1. Mature Teratoma With the Area of Thyroid Tissue.

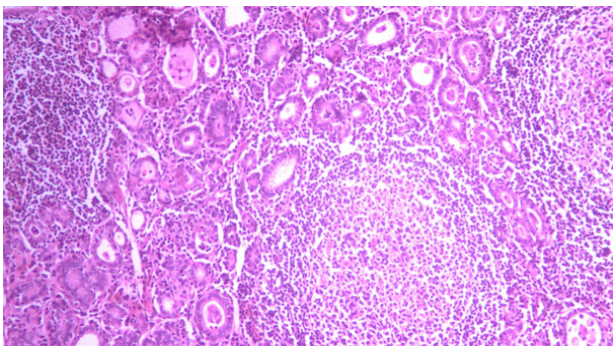

Figure 2. Lymphocytic Thyroiditis With Lymphoid Follicle Formation.

Full recovery and remission after surgery are commonly noted in patients with no ascites or pleural effusion. In benign SO without metastasis, the prognosis is good and remission occurs mostly during the 4 months to 3 years of the follow-up.

In the recent report, the general appearance after the surgery was normal and no evidence of metastasis was found. However, regarding the thyroid function, the symptoms of subclinical Hashimoto's hypothyroidism and increased thyroglobulin antibody levels were noted and levothyroxine was administered accordingly, although this problem has been reported in previous reports, other reports mentioned normal anti-thyroid antibody with clear signs of hypothyroidism. (8, 9, 17, 18).

In a report, remission was noted after 11 months of follow-up and treatment (8) and in another case regarding a malignant SO with peritoneal involvement and multiple papillary thyroid cancer, the remission was observed after 104 months of follow-up (14). The probability of malignant SO should be considered in patients with a history of autoimmune thyroiditis and surgery for benign SO because the prognosis is difficult and may be performed years after the initial diagnosis (14). More importantly, patients should be accurately monitored for any signs of hypothyroidism after the surgery because only active thyroid tissues may exist in the ovary.

Conclusion

Overall, the investigators recommend considering SO in women with ovarian mass and impaired thyroid functions.

Conflict of Interest Disclosures

No potential conflict of interests was reported relevant to this study.

Acknowledgements

We would like to thank our patient for permitting us to publish his case report.

Ethical Statement

This study was approved by the Ethics Committee of Guilan University of Medical Sciences, Rasht, Iran (Ethics code: IR.GUMS.REC.1398.424).

Authors' Contribution

All authors contributed according to the planning and writing of this case report.

Funding/Support

This study was not sponsored.

Informed Consent

Obtained.

References

1. Dunzendorfer T, deLas Morenas A, Kalir T, Levin RM. Struma ovarii and hyperthyroidism. Thyroid. 1999;9(5):499-502. doi: 10.1089/thy.1999.9.499.
2. Kondi-Pafiti A, Mavrigiannaki P, Grigoriadis C, Kontogianni-Katsarou K, Mellou A, Kleanthis CK, et al. Monodermal teratomas (struma ovarii). Clinicopathological characteristics of 11 cases and literature review. Eur J Gynaecol Oncol. 2011;32(6):657-9.
3. Yoo SC, Chang KH, Lyu MO, Chang SJ, Ryu HS, Kim HS. Clinical characteristics of struma ovarii. J Gynecol Oncol. 2008;19(2):135-8. doi: 10.3802/jgo.2008.19.2.135.
4. Kraemer B, Grischke EM, Staebler A, Hirides P, Rothmund R. Laparoscopic excision of malignant struma ovarii and 1 year follow-up without further treatment. Fertil Steril. 2011;95(6):2124.e9-12. doi: 10.1016/j.fertnstert.2010.12.047.
5. DeSimone CP, Lele SM, Modesitt SC. Malignant struma ovarii: a case report and analysis of cases reported in the literature with focus on survival and I131 therapy. Gynecol Oncol. 2003;89(3):543-8. doi: 10.1016/s0090-8258(03)00141-0.
6. Yassa L, Sadow P, Marqusee E. Malignant struma ovarii. Nat Clin Pract Endocrinol Metab. 2008;4(8):469-72. doi: 10.1038/ncpendmet0887.
7. Young RH. New and unusual aspects of ovarian germ cell tumors. Am J Surg Pathol. 1993;17(12):1210-24. doi: 10.1097/00000478-199312000-00002.
8. Berendt-Obolonczyk M, Siekierska-Hellmann M, Wojtylak S, Obolonczyk L, Sworczak K. From struma ovarii to Hashimoto disease--an unusual diagnosis of primary hypothyroidism: case report. Gynecol Endocrinol. 2012;28(1):43-5. doi: 10.3109/09513590.2011.588750.
9. Lupi I, Fessehatsion R, Manca A, Cossu-Rocca P, Martino E, Macchia E. Hashimoto's thyroiditis in a benign cystic teratoma of the ovary: case report and literature review. Gynecol Endocrinol. 2012;28(1):39-42. doi: 10.3109/09513590.2011.579659.
10. Kano H, Inoue M, Nishino T, Yoshimoto Y, Arima R. Malignant struma ovarii with Graves' disease. Gynecol Oncol. 2000;79(3):508-10. doi: 10.1006/gyno.2000.5966.
11. Savelli L, Testa AC, Timmerman D, Paladini D, Ljungberg O, Valentin L. Imaging of gynecological disease (4): clinical and ultrasound characteristics of struma ovarii. Ultrasound Obstet Gynecol. 2008;32(2):210-9. doi: 10.1002/uog.5396.
12. Outwater EK, Siegelman ES, Hunt JL. Ovarian teratomas: tumor types and imaging characteristics. Radiographics. 2001;21(2):475-90. doi: 10.1148/radiographics.21.2.g01mr09475.
13. Watson AA. Histological Hashimoto's disease in a benign cystic ovarian teratoma. J Clin Pathol. 1972;25(3):240-2. doi: 10.1136/jcp.25.3.240.
14. Russo M, Marturano I, Masucci R, Caruso M, Fornito MC, Tumino D, et al. Metastatic malignant struma ovarii with coexistence of Hashimoto's thyroiditis. Endocrinol Diabetes Metab Case Rep. 2016:160030. doi: 10.1530/edm-16-0030.
15. 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(3):112-4. doi: 10.1590/s0041-87812002000300006.
16. Ezon I, Zilbert N, Pinkney L, Wei JJ, Malik R, Nadler EP. A large struma ovarii tumor removed via laparoscopy in a 16-year-old adolescent. J Pediatr Surg. 2007;42(8):E19-22. doi: 10.1016/j.jpedsurg.2007.05.003.
17. Hosseini A, Moeini A. Clinical Finding and Thyroid Function in Women with Struma Ovarii. J Cancer Res. 2013;2013:717584. doi: 10.1155/2013/717584.
18. Manikkam B. Benign Tumor Mimicking Malignancy. J SAFOG. 2018;10(4S1):340-2.