CASE REPORT

Metastatic Follicular Variant of Papillary Thyroid Carcinoma Arising From Struma Ovarii: A Case Report

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ABSTRACT

The follicular variant of papillary thyroid carcinoma arising from struma ovarii rarely occurs, usually asymptomatic with a risk of distant metastasis. We present a case of a 37-year-old female diagnosed with malignant struma ovarii, which has histopathologic characteristics of a follicular variant of papillary thyroid carcinoma. She was discovered to have a left ovarian cyst during her second pregnancy, and laparoscopic cystectomy was performed for the left multiloculated follicular cyst. The histopathological examination came back as struma ovarii. During a subsequent pregnancy, she underwent open left salpingo-oophorectomy for a twisted left ovarian cyst. The histopathology of her left ovary revealed a follicular type of papillary thyroid carcinoma arising from struma ovarii. Following that, total thyroidectomy was performed and histopathology revealed papillary thyroid microcarcinoma. Computed tomography (CT) scan staging showed bilateral lung metastases. Subsequently, she underwent radioactive iodine therapy and survived without recurrence after six years of follow-up.

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INTRODUCTION

Struma ovarii mainly consists of mature thyroid tissue and accounts for more than half of the total tissue. The classification of struma ovarii as benign or malignant depends on the histopathologic feature. Malignant struma ovarii is a rare tumor, with 70% of cases being papillary carcinoma and only 30% being follicular carcinoma (1). The disease is rare, making treatment difficult. We present here the uncommon case of the metastatic follicular variant of papillary thyroid carcinoma arising from struma ovarii with good response to surgery and radioactive iodine therapy without recurrence after six years of follow-up.

CASE REPORT

A 37-year-old female was incidentally diagnosed with a left ovarian cyst during the first trimester of her second pregnancy. Transabdominal ultrasound results showed a multiloculated cyst over the left ovary which measured 7x5cm with the solid component. The right ovary was normal. She was asymptomatic and did not have abnormal vaginal bleeding. She underwent laparoscopic cystectomy for the left multiloculated follicular cyst and the histopathological examination came back as struma ovarii.

She had a recurrent ovarian cyst during her third pregnancy and underwent open left salpingo-oophorectomy for a twisted left ovarian cyst, which was complicated with intraabdominal bleeding and complete miscarriage. The histopathological examination of the left ovary demonstrated the follicular variant of papillary thyroid carcinoma arising from struma ovarii (Fig. 1). The tumor marker showed elevated cancer antigen 125 (CA 125), 471 U/mL (<35 U/mL), normal carcinoembryonic antigen, 0.6 ug/L (<3 ug/L) and elevated alpha-fetoprotein, 13.9 IU/mL (<3.48 IU/mL). CT scan staging reported diffuse lung metastasis with a complex right ovarian cyst (Fig. 2). Laparoscopic cystectomy for the right ovarian cyst was performed. The histopathology result of the right ovary came back as a dermoid cyst.

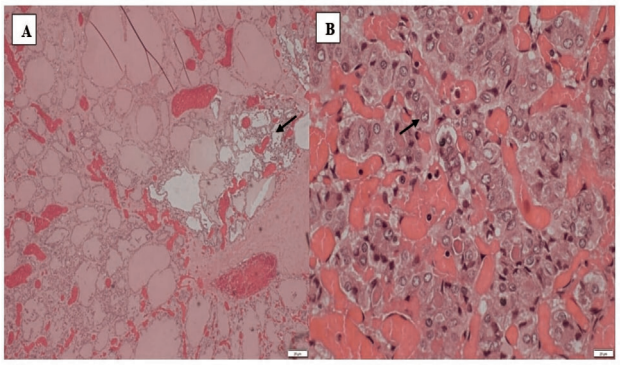

Fig. 1: Microscopic appearance of the ovarian tissue. (A) Neoplastic follicular cells with papillary thyroid carcinoma nuclear features. (B) Nuclear features of papillary thyroid carcinoma: nuclear overlapping, ground-glass nuclei, and enlarged oval-shaped nuclei (black arrow).

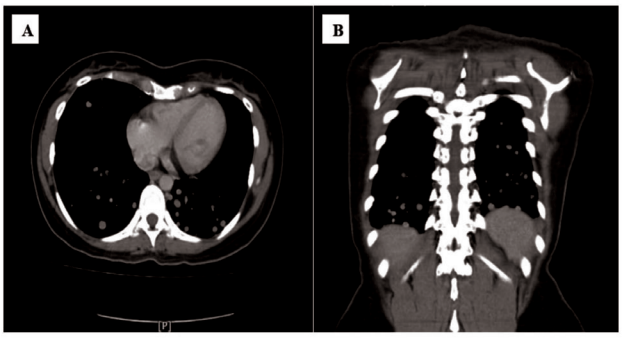

Fig. 2: CT staging shows diffuse lung metastasis. A: axial view, B: coronal view. Numerous lung nodules seen scattered throughout both lungs.

She was clinically and biochemically euthyroid. The ultrasound results of the thyroid showed a heterogeneous lesion within the left thyroid gland and a small nodule at the superior pole of the right thyroid lobe. Following total thyroidectomy, the histopathological analysis of the thyroid gland revealed papillary thyroid microcarcinoma. She was referred to the radionuclear team and received 150mCi I-131 doses of radioactive iodine (RAI) therapy. Following treatment, a whole-body thyroid scan (WBS) showed iodine avid disease in the mediastinum, lungs, abdomen, pelvis, and right femur. Subsequently, she underwent five times of RAI therapy of 150mCi I-131, with the latest WBS showing no more uptake. She is currently on thyroxine suppression dose therapy and calcium supplement.

DISCUSSION

More than half of the tissue in a struma ovarii consists of the thyroid tissue, which is also known as ovarian teratoma. Although struma ovarii is mostly benign, 5% -10% is malignant. It can also be histologically identified as differentiated thyroid carcinoma (2). The frequent type of malignancy that occurs in stuma ovarii is papillary carcinoma, not the follicular type. As for thyroid gland carcinoma, typical follicular carcinomas can metastasize to distant sites including the abdominal cavity, liver, bone, lungs, and opposite ovary (1).

Although malignant struma ovarii (MSO) can affect women at any age, it most frequently occurs in their fourth to sixth decades of life (3). Most patients with MSO are asymptomatic, and often clinically presents as an ovarian tumor (4). The most typical presentation includes abnormal vaginal bleeding, ascites, pelvic mass, and abdominal pain. 5-8% of struma ovarii cases will present with thyroid dysfunction symptoms like thyrotoxicosis and hyperthyroidism. Usually, the tumor is unintentionally found during pelvic or abdominal ultrasound, CT scanning, or unrelated surgery (1). In this case, the patient was asymptomatic and incidentally diagnosed after a surgical operation for the twisted left ovarian cyst.

The follicular form of MSO is a rare tumor. The proliferation of cells grouped in trabecular and follicular patterns was its defining feature. In this case, the ovary's histopathological characteristics revealed predominantly thyroid follicles showing extensive hemorrhage and degenerative changes. The follicles vary in size, containing colloids with some crowded areas. A simple cuboidal epithelium with nuclei positioned basally lines the majority of the follicles.

Approximately 5% of MSO cases will eventually develop distant metastases, which rarely occurs (4). The metastases can spread via blood and lymphatic channels (1). The main site of metastasis is the intraabdominal cavity, which also includes the fallopian tubes, peritoneum, contralateral ovary, and paraaortic lymph nodes. The brain, bones, lungs, and liver can all be affected by blood-borne metastases (1). In the present patient, distant metastasis occurs in the mediastinum, lungs, abdomen, pelvis, and right femur.

The primary site of distant metastasis in this case is the ovarian lesion. The foci of papillary thyroid microcarcinoma in the thyroid gland measured about 10mm. Hence, it is considered as papillary thyroid microcarcinoma, which is theoretically not commonly metastasis. Approximately 90% of papillary thyroid microcarcinomas do not grow, and almost 96% of the tumors do not spread to the lymph node over the course of ten years (5). On the other hand, there were multiple foci of varying sizes of follicular variant of papillary thyroid carcinoma in the left struma ovarii, which is more likely to be the primary malignant lesion.

As the follicular variant of malignant struma ovarii rarely occurs, the treatment is also challenging and difficult. There are limited data on the optimal diagnostic standards, course of treatment, and duration of surveillance for this disease. In postmenopausal or premenopausal women who have completed childbearing, treatment includes bilateral salpingo-oophorectomy with omentectomy and total abdominal hysterectomy. Women who want

to preserve their fertility are recommended to have unilateral cystectomy or salpingo-oophorectomy. Aggressive treatment for metastatic struma ovarii is required which comprises total thyroidectomy and adjuvant radioactive iodine treatment (1). Radioactive iodine aims to destroy the thyroid cancer metastatic foci and ablation of thyroid remnants. Whole-body scanning and serum thyroglobulin level measurement are used for disease monitoring.

In this case, after total thyroidectomy and radioactive therapy, the tumor spread to the mediastinum, lungs, abdomen, pelvis, and right femur. No more metastasis was detected with repeated radiotherapy ablation treatment. The serum thyroglobulin level after 1 year post-radiotherapy ablation treatment was 2.7 ng/mL (<0.1ng/mL) with a negative thyroglobulin antibody. The serum thyroglobulin levels should be continuously monitored to identify tumor recurrence (3).

CONCLUSION

The diagnosis and management of the follicular variant of papillary thyroid carcinoma arising from struma ovarii remain challenging. Treatment choices must be decided on an individual basis based on suspicion and histological evaluation due to the rare nature of the disease. A close observation for more than 10 years is recommended to identify the disease recurrence (3).

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