

Concurrent Intrathyroidal Follicular Variant of Papillary Thyroid Carcinoma with Malignant Struma Ovarii Presenting 12 Years After Initial Diagnosis

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Malignancy in struma ovarii is rare and observed in only 5%–10% of patients. Here, we present a patient with malignant struma ovarii and coexisting intrathyroidal papillary thyroid carcinoma, with recurrence (large pouch-of-Douglas mass) and metastases (bilateral pulmonary and iliac nodal metastases) presenting 12 y after surgery. The notable features in this case were a concurrent intrathyroidal follicular variant of papillary carcinoma; the highly functioning nature of the malignant lesions, characterized by a low level of thyroid-stimulating hormone even without thyroxine suppression; and the low-grade ^{18}F -FDG avidity of these lesions, consistent with their well-differentiated nature. With the adoption of a multimodality approach (surgery, radioiodine scintigraphic evaluation, and multiple radioiodine therapies), the patient showed a progressive decrease in the functionality of the disease, prolonged progression-free survival, and a good quality of life with symptom-free status at 5 y.

Key Words: malignant struma ovarii; follicular variant of papillary thyroid carcinoma; quality of life; radioiodine therapy; ^{131}I

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Struma ovarii is a rare, specialized type of ovarian teratoma constituting 2%–5% of all ovarian teratomas and 1% of all ovarian neoplasms (1). Struma ovarii is defined as a teratoma that substantially comprises mature thyroid tissue ($\geq 50\%$) (2). Approximately 5%–10% of struma ovarii are malignant, with papillary thyroid carcinoma being the most common histopathologic type, followed by follicular carcinoma (3). Patients most commonly present with pelvic mass symptoms such as ascites or pain (4). The most common sites of metastases are the peritoneum, mesentery, and omentum, followed by

the liver, contralateral ovary, bone, and lung (5). We describe a case of simultaneous intrathyroidal thyroid cancer with follicular variant papillary carcinoma of the thyroid arising in struma ovarii, which recurred after 12 y.

CASE REPORT

A 52-y-old woman with histologically confirmed struma ovarii had undergone total abdominal hysterectomy with bilateral salpingo-oophorectomy 12 y previously and presented recently with lower abdominal pain and irregular bowel habits. Contrast-enhanced CT (thorax, abdomen, and pelvis) revealed a large lesion (7.5×6.1 cm) in the pouch of Douglas, soft-tissue deposits in the right internal iliac region, and multiple well-defined nodules bilaterally in the lung parenchyma. Histopathologic examination of the pelvic mass showed mature thyroid tissue (predominantly follicular variant papillary carcinoma of the thyroid) with stroma, whereas that of the lung nodules showed metastatic carcinoma of the thyroid with a follicular pattern. The pelvic mass was reported as recurrence of struma ovarii. Ultrasonography of the neck showed multiple suggestive hypoechoic nodules in both lobes of the thyroid,

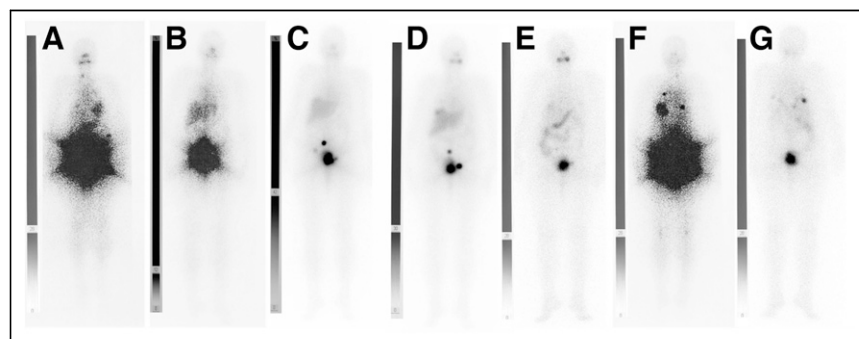


FIGURE 1. (A–E) Posttherapy ^{131}I scan (anterior images) demonstrating iodine uptake in pelvic lesion after first RAI cycle (A), second cycle (B), third cycle (C), fourth cycle (D), and fifth cycle (E), showing progressive response. (F and G) Posterior images demonstrating iodine uptake in lung nodules and pelvic lesions after first RAI cycle (F) and after fifth cycle (G).

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with a score of 4 on the Thyroid Imaging Reporting and Data System, which were proven to be papillary thyroid carcinoma (Bethesda category VI) on fine-needle aspiration cytology. The patient underwent total thyroidectomy, with histopathologic

TABLE 1

Thyroglobulin and Thyroid-Stimulating Hormone Values over Course of Treatment from Baseline to Last Follow-up

Time	Thyroglobulin (ng/mL)	Thyroid-stimulating hormone (μ IU/mL)
Thyroid-stimulating hormone stimulated		
Baseline (before first RAI cycle)	>300	1.67
6 mo after first RAI cycle	>300	13.98
1 y after second RAI cycle	300	58
1 y after third RAI cycle	288	52
1 y after fourth RAI cycle/ before fifth RAI cycle	269	46.6
Thyroid-stimulating hormone suppressed		
Recent follow-up (6 mo after fifth RAI cycle)	75	0.008

examination indicative of follicular variant papillary carcinoma of the thyroid. Diagnostic ^{131}I planar scintigraphy was performed with 3.7 MBq of ^{131}I 4 wk after surgery and demonstrated bilateral radioiodine (RAI) uptake in the lungs and the pelvic mass. After treatment with 7,622 MBq (206 mCi) of RAI, a scan acquired at the time of discharge (Fig. 1) showed iodine-avid nodules bilaterally in the lungs and intense uptake in the pelvic mass. The patient received 5 cycles of RAI therapy, with the second cycle being administered 6 mo after the first cycle and the third, fourth, and fifth cycles being administered around 12–13 mo after the second, third, and fourth cycles, respectively. The RAI activity per cycle ranged from 6,401 MBq (173 mCi) to 7,955 MBq (215 mCi), with a cumulative dose of 35,594 MBq (962 mCi); the last cycle of therapy was 12 mo previously. After 2 cycles of RAI therapy, the thyroid-stimulating hormone level increased to 58 μ IU/mL (it had been 1.67 μ IU/mL at baseline), suggestive of a good response by the hyperfunctioning metastases. The patient's stimulated serum thyroglobulin was more than 300 ng/mL at baseline and 269 ng/mL after 4 RAI cycles, whereas the

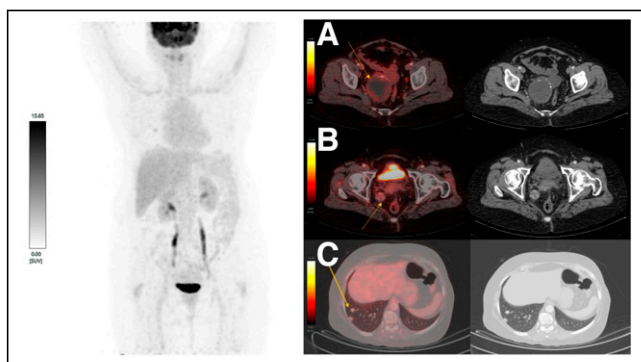


FIGURE 2. Maximum-intensity projection image of ^{18}F -FDG PET/CT scan showing very low-grade ^{18}F -FDG uptake (arrows) in pelvic mass (A), right internal iliac deposit (B) and lung nodules (C). (A–C) Left-side images are fused PET/CT axial images, and right-side images are CT axial images.

suppressed serum thyroglobulin was 75 ng/mL at 6 mo after the fifth RAI cycle (Table 1). Antithyroglobulin antibody was negative on all occasions.

^{18}F -FDG PET/CT (Fig. 2) acquired at the time of the last follow-up (6 mo previously) showed a low-grade ^{18}F -FDG-avid mass in the pouch of Douglas ($7.6 \times 7 \times 7.9$ cm; SUV_{max} , 4.4), the right internal iliac deposit, and bilaterally in the metastatic lung nodules. Stable disease was achieved after RAI therapy. The total follow-up period was 5 y since the initial presentation. The patient was subsequently observed on thyroxine suppression and follow-up. She is doing well, with an Eastern Cooperative Oncology Group performance score of 1.

DISCUSSION

Concurrent struma ovarii with intrathyroidal thyroid carcinoma has been rarely reported and has no defined guidelines for management. DeSimone et al. (5) reviewed 24 cases of malignant struma ovarii and observed that patients treated with ^{131}I as adjuvant therapy showed no recurrence and that patients who developed recurrence later showed a complete response to radioablation. Multimodality treatment with surgical resection followed by RAI scanning and adjuvant RAI therapy is recommended as a first-line approach for malignant struma ovarii (2,5). ^{131}I scintigraphy has been used to detect residual or recurrent disease and evaluation for treatment with ^{131}I (2). In our case, the patient was successfully managed by total thyroidectomy followed by RAI scanning and therapy. In patients with metastatic disease, RAI ablation and thyroid-stimulating hormone suppression are associated with increased disease-free survival and reduced recurrence rates.

CONCLUSION

A rare case of concurrent intrathyroidal follicular variant papillary carcinoma of the thyroid with malignant struma ovarii and RAI-avid metastatic disease was successfully managed with total thyroidectomy and RAI therapy, demonstrating a progressive decrease in the functionality of the disease, prolonged progression-free survival, and a good, symptom-free quality of life.

DISCLOSURE

No potential conflict of interest relevant to this article was reported.

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