Follicular carcinoma in a functioning struma ovarii

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Summary

We describe a case of follicular carcinoma in a functioning struma ovarii, which presented as an ovarian mass in a patient who had undergone a near-total thyroidectomy for a benign lesion. She underwent bilateral salpingo-oophorectomy and received radiotherapy and L-thyroxine treatment with no evidence of metastases in 4 years follow-up.

Keywords: struma ovarii; follicular carcinoma

Struma ovarii is an uncommon tumour, and its malignant transformation is rare.¹ In most instances, diagnosis of malignant struma ovarii is made postoperatively.² We report a patient who had undergone near-total thyroidectomy many years earlier for haemorrhage into a thyroid nodule, who presented with an ovarian mass which was diagnosed as a functioning struma with follicular carcinoma, recognised during hormone replacement therapy (HRT).

Case report

A 47-year-old woman presented with an euthyroid solitary nodule in June 1987. The nodule was hypoechoic with poor ¹³¹I-uptake on scan and had features of follicular neoplasm on fine needle aspiration cytology. Her serum T₃ and T₄ were normal (1 ng/ml and 100 ng/ml, respectively). She had a near-total thyroidectomy for haemorrhage into the nodule. However, the detailed histology of the nodule excluded malignancy. She had received 150 µg L-thyroxine daily until May 1990. Since then she had remained euthyroid with no palpable goitre and normal serum T₃ and T₄

levels. In March 1994, she complained of irregular cycles and experienced hot flushes. She was put on HRT with conjugated oestrogen and medroxyprogesterone after excluding gynaecologic abnormality by vaginal PAP smear and pelvic ultrasound.

Her hot flushes decreased, but after 6 months she had intermenstrual bleeding. A tender right fornix and 5 × 6 cm firm mobile mass was found on per vaginal examination and confirmed by pelvic ultrasound. At laparotomy, a 7 × 6 cm right ovarian mass was seen with variegated appearance and multiple solid and cystic areas with an intact surface. There was ascites with no visible peritoneal seedlings. Bilateral salpingo-oophorectomy and hysterectomy were performed. Ascitic fluid was negative for malignant cells. Histopathology revealed near-complete transformation of ovarian tissue into sheets of cells arranged in a follicular pattern with thin colloid separated by fibrocollagenous tissue. The follicular cells showed nuclear pleomorphism, mitotic activity and were invading its capsule; there was no vascular invasion. These findings were consistent with a diagnosis of follicular carcinoma in struma ovarii. No other teratomatous elements were seen (figures 1 and 2). Oestrogen and progesterone receptor status by peroxidase anti-peroxidase immunochemistry after microwave retrieval were found to be negative. The left ovary was normal. An ¹³¹I whole body scan after 6 weeks of laparotomy revealed uniform uptake in residual thyroid tissue (2.4% at 24 h) and no tracer uptake elsewhere. Her unsuppressed serum thyroglobulin level was 4.5 ng/ml (normal < 10 ng/ml). She had low serum T₃ and T₄ (0.55 and 40 ng/ml, respectively) and an elevated thyroid-stimulating hormone

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Figure 1 The section shows sheets of thyroid follicular cells with thin colloid (H&E, orig × 256)

Figure 2 The cancerous cells invading into the capsular lymphatics (H&E, orig × 256)

(TSH) of 40.8 $\mu\text{U/ml}$, indicating hypothyroidism. She received 4500 rads external beam radiotherapy to the pelvic region in view of the capsular invasion and ascites, together with L-thyroxine therapy.

She is currently well with no evidence of metastases on total body scan at 4 years post-operative follow-up

Discussion

'Struma ovarii' is a slow-growing ovarian neoplasm, with thyroid tissue as its only or the predominant (>50%) constituent.³ The ovarian thyroid is histologically and functionally identical to cervical thyroid.⁴ The tumour is a highly specialised subclass of benign cystic teratoma¹; 95% of them remain benign while the remainder undergo malignant transformation, with peak frequency during the fifth decade of life. The left ovary is more frequently involved than the right and in 6% of instances struma ovarii are bilateral.¹

Struma ovarii often present with abdominal mass, lower abdominal pain, ascites and, uncommonly, hyperthyroidism (5%).³ Past or concomitant thyroid enlargement has been described with struma ovarii (18%) which may cause difficulty in its diagnosis.³ Retrospectively, the fact that this patient remained euthyroid without L-thyroxine replacement for 4 years, could have given a clue to the presence of a functioning struma ovarii. This was further substantiated when the patient became hypothyroid (TSH 40.8 $\mu\text{U/ml}$) promptly after the removal of the ovarian mass. An ¹³¹I whole body scan prior to exploratory laparotomy would have picked up the lesion.

Cellular atypia, nuclear pleomorphism, vascular and/or capsular invasion and distant metastases are definite clues to malignant transformation of struma ovarii.^{4,5} Nuclear pleomorphism, mitotic activity and capsular invasion in our patient suggested the malignant

Learning points

- struma ovarii is an uncommon cause of hyperthyroidism
- toxicosis or euthyroidism in a patient with near-total thyroidectomy should raise the suspicion of functioning struma ovarii
- an ¹³¹I whole body scan prior to exploratory laparotomy would have established the diagnosis of struma ovarii
- treatment modalities include surgery and/or radioiodine ablation and L-thyroxine in TSH-suppressive doses

nature of the tumour. Capsular invasion carries a poor prognosis.^{4,5} The majority of malignant struma present as follicular carcinoma, while papillary, anaplastic, and Hurthle cell carcinoma have also been described.⁵

Recognition of an ovarian mass as a struma ovarii during the course of oestrogen–progesterone therapy is an interesting event. Oestrogen modulating TSH secretion and/or TSH exerting a positive influence on follicular growth might have been responsible for the rapid growth of the struma ovarii.⁶ It is noteworthy that the lack of demonstration of oestrogen and progesterone receptors in our case can be explained by the complete transformation of ovarian tissue into struma, as normal thyroid follicular cells do not express oestrogen receptors.

Therapeutic modalities include total abdominal hysterectomy with bilateral salpingo-oophorectomy, thyroid ablation (radioiodine or surgery) followed by thyroxine-suppressive treatment. Chemotherapy and radiotherapy have been used for recurrent metastatic struma ovarii and anaplastic carcinoma which do not concentrate radioiodine.^{1,7}

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