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CASE REPORT

Papillary type thyroid carcinoma in an ovarian struma

J. P. O'Neill · P. Burns · J. Kinsella

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Abstract
Background Struma Ovarii are mature teratomas. In rare circumstances thyroid tissue is found as part of the histopathological makeup. Malignant transformation may occur in 1–2% of these rare cases.
Aim To report a rare case of malignant thyroid carcinoma within a struma ovarii.
Result A 22-year-old lady presented with a right pelvic mass. A right-sided laparoscopic salpingo-oophrectomy was performed which revealed a malignant struma ovarii neoplasm. A total thyroidectomy was performed with adjuvant I^{131} therapy.
Conclusion This is a rare tumour which necessitates surgical extirpation and removal of a normal thyroid gland to facilitate thyroglobulin monitoring.

Keywords Struma ovarii · Papillary thyroid carcinoma · Oophrectomy · Teratoma

Introduction

Struma ovarii is a form of specialised mature teratoma which originates from a three germ cell layers. These are usually unilateral on presentation. Mature thyroid tissue in an ovarian teratoma is reported in approximately 2% of cases. Malignant transformation occurs in 1–2% of patients, normally in the older, post menopausal populations. Treatment involves pelvic tumour extirpation and adjuvant radioiodine (I^{131}) therapy. Tumour surveillance with thyroglobulin necessitates total thyroidectomy.

J. P. O'Neill (✉) · P. Burns · J. Kinsella
The Department of Otolaryngology, Head and Neck Surgery, St James Hospital, Dublin, Ireland
e-mail: joneill@rcsi.ie; drjponeill@hotmail.com

P. Burns
e-mail: pauljburns@eircom.net

J. Kinsella
e-mail: johnkinsella@eircom.net

Case report

We report a 22-year-old lady who presented with an abdominal mass and urinary compressive symptoms. The patient had no co-morbidities and no history of polycystic ovarian syndrome, prolactinoma, thyroid disease, breast or ovarian cancer. Gynaecological anamnesis reported menarche at the age of 14 years and regular menstrual cycles, parturition 1, abortion 0. Examination revealed a fixed, tender abdomino-pelvic mass. Laboratory blood investigations were normal including all hormone profiles including thyroid function and tumour markers were normal.

A normal uterus was confirmed on ultrasonography. Magnetic Resonance Imaging (MRI) reported a right-sided multi-cystic ovarian mass arising from the right hemi pelvis with fat and nodular components. The size was estimated at $150 \times 70 \times 160$ mm. A moderate right-sided secondary hydronephrosis was noted. This mass was found to be compressing the Inferior Vena Cava (IVC) and right common iliac vein and a large volume of free fluid was noted lying dependently in the pelvis.

A right-sided laparoscopic salpingo-oophrectomy was performed which revealed a malignant struma ovarii neoplasm. This was found to include typical teratomatous mature components of sebaceous elements and cartilage. Within the teratoma, epithelial origins were also observed, in particular an unencapsulated neoplasm with papillae

consisting of neoplastic epithelium overlying fibrovascular stalks. Nuclei had an empty ground-glass appearance with characteristic nuclear grooves, pseudoinclusions and calcific concretions with a circular laminated appearance (Fig. 1). Immuno-histochemical expression of Thyroglobulin (Fig. 2) and Thyroid Transcription Factor (Fig. 3) was also confirmed. The teratoma was largely replaced by a malignant papillary thyroid carcinoma.

A thyroidectomy was performed to facilitate thyroglobulin monitoring. The thyroid gland was normal with no evidence of malignancy. She then underwent adjuvant radioactive iodine by administration of 104 mCi (131) I. No scintigraphic evidence of functioning thyroid tissue was noted in the thorax, abdomen or pelvis. A repeat scan 9 days' post therapy failed to reveal a focal area of abnormal isotope uptake.

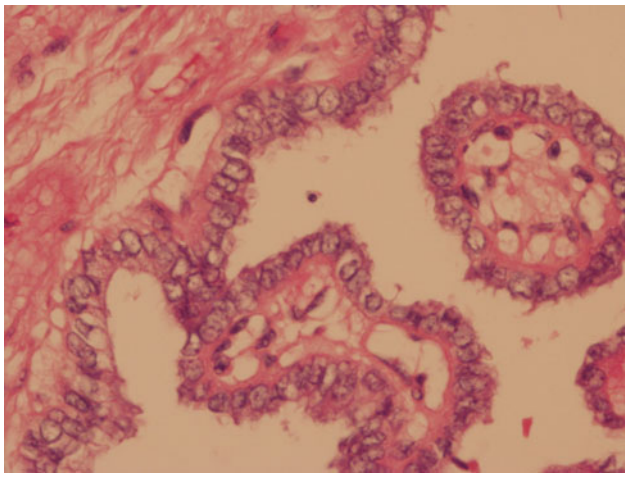

Fig. 1 Malignant struma ovarii tumour with papillary thyroid carcinoma in situ

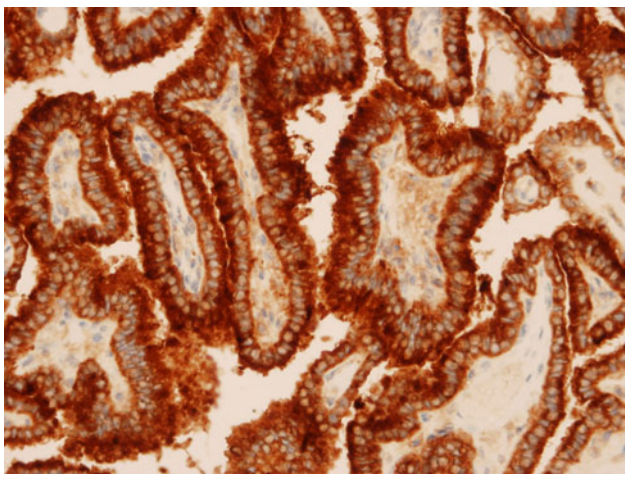

Fig. 2 Struma ovarii stained positive for Thyroglobulin

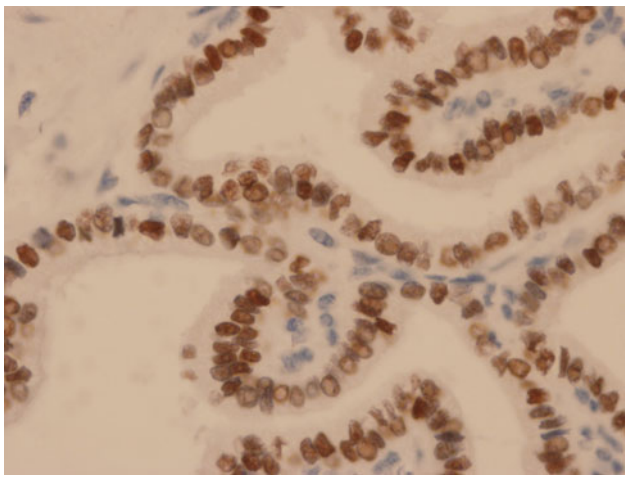

Fig. 3 Struma ovarii stained positive for Thyroid Transcription Factor

Discussion

Struma Ovarii tumours are mainly benign unilateral lesions and rarely have a malignant focus. They are monomeric and highly specialised teratomas accounting for less than 5% of mature teratomas [1]. Struma ovarii is defined by the presence of an ovarian tumour containing thyroid tissue as the predominant cell type and was first described at the turn of the century by Professor Ludwig Pick [2].

Preoperative radiological diagnosis is complicated by a lack of specificity on the part of MRI, Computer Tomography (CT) and Ultrasonography. The tumours are at best interpreted as adnexal masses with solid and cystic components. In particular it is difficult to distinguish between struma ovarii and dermoids on sonographic appearance; however, overall CT and MRI can assist differentiation [3, 4]. Preoperative scintigraphy with I^{131} has been shown to outline active thyroid tissue activity within the Pelvis [5].

Pseudo-Meigs Syndrome is a rare association of the Struma ovarii tumour with abdominal ascites and pleural effusion [6]. Strumal tissue is not normally functionally active; however, cases associated with thyrotoxicosis have been reported and can be due to autoimmune stimulation of the normal thyroid gland.

Malignant transformation of the thyroid tissue can be papillary, follicular or of mixed pattern, and it can include elements of carcinoid, melanoma, mucinous cystadenocarcinoma or Brenner tumour.

Surgical management is the mainstay of treatment for this condition. To allow for thyroglobulin post operative estimation as an oncology marker, total thyroidectomy was performed on our patient. This allows for correct post-operative monitoring for differentiated thyroid malignancy. It also opens the patient to radioactive I^{131} therapeutic

strategy. The thyroid gland was found to be normal with no evidence of malignancy.

The patient then underwent adjuvant radioactive iodine by administration of 104 mCi (131) I. No scintigraphic evidence of functioning thyroid tissue was noted in the thorax, abdomen or pelvis. A repeat scan 9 days post therapy showed no focal isotope uptake. Even in malignant cases, adjuvant iodine-131 ablation with surgical extirpation has proven curative. Recurrences may be detected using iodine-131 scanning, and repeat iodine radioablation can lead to extended disease-free survival.

Histopathological analysis including microscopy and immunohistochemistry of multiple tumour samplings is required for definitive diagnosis [7]. Immunocytochemistry for Thyroglobulin (TGB) or Thyroid Transcription Factor (TTF-1) is also a valuable tool for preoperative fine needle aspiration biopsy and intraoperative diagnosis.

In conclusion, Struma Ovarii are challenging to diagnose both macroscopically and histologically. Although the typical presentation is that of a pelvic mass, unusual clinical manifestations such as hyperthyroidism, ascites, and Meigs' syndrome have been reported [8]. Surgery is the main treatment strategy and prognosis is excellent.

Conflict of interest statement None.

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