Thyroid type papillary carcinoma arising in a mature teratoma

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Abstract Struma ovary is a monodermal ovarian teratoma which contains mature thyroid tissue. We present an unusual case of papillary carcinoma arising in struma ovarii. A 52 year old post menopausal lady presented with a large right adnexal mass preoperatively diagnosed as a dermoid cyst. She underwent staging laparotomy and the histopathology report showed follicular variant of papillary carcinoma thyroid arising in a mature teratoma. She subsequently underwent total thyroidectomy to enable follow up with serum thyroglobulin and iodine scan. On follow up there is no evidence of disease recurrence.

Keywords Struma ovary · Thyroid carcinoma · Ovarian teratoma

Introduction

Struma ovary is a monodermal ovarian teratoma which contains mature thyroid tissue. These tumours may secrete thyroxine hormone and produce symptoms of hyperthyroidism. They are usually benign tumours although occasionally the thyroid tissue shows malignant transformation. The incidence is from 1 to 3 % of all ovarian teratomas. (1).

Malignant struma ovary is usually diagnosed post operatively based on histopathologic findings. Patients are usually asymptomatic with pelvic mass and pain being the common

presenting symptoms. 5–8 % of patients will have clinical hyperthyroidism (2).

Here we present an unusual case of papillary carcinoma arising in a teratoma.

Case

A 52 year old post menopausal female presented with abdominal distension and mass abdomen.

On examination she had a 15×15 cm right adnexal mass in the right iliac fossa and hypochondrium arising from the pelvis. Per vaginal examination revealed a right adnexal mass of about 15×15 cm. The cervix was normal. The uterus was not felt separate from the adnexal mass. There was no ascites or deposits in the rectouterine pouch. She showed no features of hyperthyroidism.

She was evaluated by CT scan abdomen which showed a large heterogenous mass with enhancing solid and cystic areas on the right side of the pelvis with the solid component measuring 9×7 cm and cystic component measuring 8×6 cm, suggestive of a right ovarian dermoid cyst.

Her serum tumour markers were normal - CA 125 was 28 ng/mL, S CEA 2.3 ng/mL. Thyroid function test done for preoperative evaluation was normal. (T3 – 2.46 ng/ml, T4 – 9.88 ng/mL, TSH – 0.67 mIU/ml).

She was taken up for staging laparotomy with intraoperative frozen study.

During laparotomy there was a $16 \times 13 \times 8$ cm bosselated highly vascular right ovarian mass adherent to the uterus, appendix and omentum (Fig. 1). There was a 3×2 cm deposit on the inferior surface of the liver. Excision of the right ovarian mass and total abdominal hysterectomy with omentectomy, appendectomy and excision of the omental deposit was done. Frozen section analysis of the ovarian mass revealed teratoma with predominant thyroid tissue.

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Fig. 1 right ovarian tumour with solid and cystic areas

Pathological findings

The ovariectomy specimen measured 15×10×9 cm with a bosselated surface. Cut section had a variegated appearance with solid and cystic areas. The cystic area was filled with putaceous material and hair. The solid area was tan colored with mucinous areas. Microscopic examination revealed thyroid follicles lined by cells with nuclear grooving, eosinophilic inclusions (Fig. 2, 3).

Pathologic diagnosis was follicular variant of papillary carcinoma thyroid arising in a mature teratoma (Fig. 4). The omental deposit showed a stitch granuloma from a previous post partum sterilisation.

The case was discussed in a multidisciplinary tumour board. Total thyroidectomy was advised to enable surveillance with serum Thyroglobulin and Iodine scan. An ultrasound neck showed a normal thyroid. A low dose iodine scan showed normal uptake in the thyroid bed and no abnormal uptake elsewhere. She was counselled about the need for surgery and she underwent Total thyroidectomy. The post operative period was uneventful and she has no hypoparathyroidism or recurrent laryngeal nerve palsy. Histopathology report of the thyroidectomy specimen showed a cellular nodule with focal clear cell change with adjacent lymphocytic thyroiditis.

On follow up at 1 year her S Thyroglobulin level is <0.01 ng/ml. She is on suppressive dose of Thyroxine hormone orally. An Iodine scan revealed no uptake in the neck or abdomen.

Discussion

The malignant struma ovarii was first described by Wetteland in 1956. (3) Malignancy in struma ovary is diagnosed based on histopathological criteria and guidelines for primary thyroid gland disease (4) Papillary and follicular carcinoma

Fig. 2 Section from the cyst shows partial lining by squamous epithelium (arrow) with areas of foreign body giant cell reaction.[H&Ex100]

are the common malignancies arising in the struma ovarii. Roth and Karseladze have described a new entity of follicular carcinoma called highly differentiated follicular carcinoma of ovarian origin (HDFCO), characterized by extra ovarian dissemination of thyroid elements with an innocuous appearance that histologically resembles nonneoplastic thyroid tissue. The peritoneal deposits seen in this type of tumours has been called peritoneal strumosis. (5) A case of pseudo-Meigs syndrome associated with ascites, hydrothorax, and elevated CA levels has been described in malignant struma ovarii. The associated symptoms disappeared, and the elevated CA 125 returned to within reference range postoperatively without adjuvant therapy. (6) Hyperthyroidism induced by malignant struma ovarii is likely to be mediated by a mechanism different from classic thyroid hyperthyroidism. It is postulated that struma ovarii is an autonomous hormone-secreting tumor or that the ovarian thyroid tissue is stimulated by thyroid-stimulating hormone receptor antibody. (7) Metastasis of malignant

Fig. 3 Section from the cyst in areas showing thyroid tissue with the follicles lined by cuboidal cells and some of the follicles containing colloid (arrow).(H&Ex200)

Fig. 4 High power view from solid area shows follicular variant of papillary carcinoma thyroid, with many cells showing large vesicular nucleus with nuclear grooves (arrow).[H&Ex400]

struma ovarii is seen in approximately 5 to 23 % of cases and is mainly intra-abdominal, although blood-borne metastasis can occur in the liver, lung, brain, bone, vertebra, and the contralateral ovary. (2) Osseous metastases occurred in 26 % of patients with typical follicular carcinoma and 17 % of patients with HDFCO, but only 4 % of patients with papillary carcinoma. Typical follicular carcinoma is more likely to metastasize to the lung, liver, and central nervous system; whereas papillary carcinoma has a propensity to involve the abdominal cavity and lymph nodes and occasionally the liver. (5).

Consensus on the surgical and postoperative treatment of patients with thyroid-type carcinoma arising in struma ovarii has not been definitively reached. Surgical treatment ranges from total abdominal hysterectomy, plus bilateral salpingo-oophorectomy with omentectomy, to conservative surgery, involving unilateral oophorectomy or strumectomy (cystectomy), for preservation of fertility. Adjuvant therapy includes external radiotherapy, chemotherapy and thyroid suppression. (2). Some authors recommend thyroidectomy and I131 ablation as the first-line treatment in cases with extraovarian dissemination or when preservation of fertility

is not required after surgical diagnosis, and they have found I131 therapy prevented local and distant recurrence. (8) (9).

Aggressive surgery could achieve complete resection of the tumour in our patient. Intraoperative frozen section was able to correctly diagnose struma ovary. However the malignancy hidden within the struma was revealed only on H&E staining of the fixed specimen. Being a rare condition there are no guidelines to the need for thyroidectomy in this patient. Iodine scanning and thyroglobulin monitoring for recurrence detection is more sensitive if the native thyroid is removed. Considering the low morbidity associated with thyroidectomy at our centre and the complete resection of the primary tumour in the abdomen it was decided at the multidisciplinary tumour board to offer total thyroidectomy for this patient. The excellent outcome and undetectable disease on follow up at the end of 1 year has vindicated our stance. This case is being reported for its rarity and the need for aggressive surgical management in cases.

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