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COMMUNICATION

Papillary carcinoma in struma ovarii, laparoscopic management—a case report

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Abstract Struma ovarii is an ovarian teratoma in which thyroid tissue predominates. Malignancy, either papillary or follicular type in struma ovarii is an extremely rare event. We report a case of papillary carcinoma in struma ovarii in a 27-year-old lady who was admitted with an acute pain in the abdomen. She underwent laparoscopic cystectomy, followed by laparoscopic unilateral salpingo-oophorectomy, total thyroidectomy, and iodine-131 treatment on confirmation of the diagnosis of papillary carcinoma in struma ovarii. Malignancy in struma ovarii can be managed satisfactorily with a combination of surgery and treatment options currently available for primary thyroid cancer. Papillary variant has a more favourable prognosis than its follicular counterpart.

Keywords Ovarian teratoma · Malignant struma ovarii · Thyroid papillary carcinoma

Introduction

Mature cystic teratomas account for around 40% of all ovarian neoplasms. The term struma ovarii is used for tumours composed predominantly of thyroid tissue and comprises only 1.4% of cystic teratomas. Malignancy in struma ovarii is rare and occurs only in 5–10% of cases.

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Case

A 27-year-old nulliparous woman was admitted under the general surgeons with acute pain in the lower abdomen. She did not have any significant medical or surgical history, used combined oral contraceptive pill and was in a stable relationship. On the ward, she developed pyrexia and hence started on intravenous antibiotics.

Gynaecological examination revealed generalised abdominal tenderness and guarding, a healthy cervix, tender uterus and adnexa.

Investigations Full blood count showed an elevated white cell count (13×10^9 per litre, ref range 4–11) and C-reactive protein 61 mg/l (ref range less than 5). Serum beta human chorionic gonadotropin was normal at less than1 U/l. There was no growth on culture of midstream sample of urine. The abdominal, renal and transvaginal pelvic ultrasound reported a smooth-walled bladder, normal-sized anteverted uterus with thin complete midline endometrial echo. A cystic mass with internal echoes was noted in the pouch of Douglas with a possible relation to either ovarian cyst, fallopian tube or appendix. Surrounding this and the uterus was echogenic-free fluid. The cystic structure measured 109×57mm. Both kidneys appeared normal and there was free fluid in the upper abdomen. Possible conclusion was either tubo-ovarian abscess or ruptured appendix.

Tumour markers were not obtained.

Management She underwent a diagnostic laparoscopy, and the findings were: grossly injected peritoneal cavity and the whole of the abdominal and pelvic cavity were filled with turbid fluid. The appendix, right ovary and tube were normal. There was a partially ruptured benign looking left

ovarian cyst and so a laparoscopic left cystectomy and copious irrigation with normal saline was carried out.

The postoperative recovery was good and she was discharged the next day.

Histopathology Macroscopic appearance showed several pieces of a collapsed cyst with smooth outer and inner walls. The largest piece was 9 cm in maximum dimension. Some of the pieces were covered by brownish material resembling altered blood. There were no discrete solid areas identified.

Microscopic appearance showed parts of multilocular cyst lined in most parts by flattened cuboidal cells and in several areas the lining appeared papillary. The cells were pleomorphic and hyperchromatic with ground glass nuclei and nuclear grooves (Fig. 1). Several follicles lined by follicular cells and filled with colloid were also present. Other areas were composed of dense fibrous tissue with occasional follicular cyst.

Immunoperoxidase staining was positive for thyroglobulin, thyroid transcription factor (TTF)-1. There was patchy positivity for CK34BE12.

Two further markers CK19 and HBME1 were used to distinguish between nonneoplastic and papillary carcinoma of thyroid.

Diagnosis papillary carcinoma in struma ovarii or metastatic spread from a papillary carcinoma within the thyroid. The diagnosis was confirmed after second opinion from a tertiary centre.

Further management The case was discussed at the multidisciplinary oncology meeting and the following management plan was formulated: left oophorectomy and peritoneal biopsies, ultrasonography of the thyroid to exclude primary thyroid carcinoma, radio iodine uptake scan and referral to the thyroid multidisciplinary team.

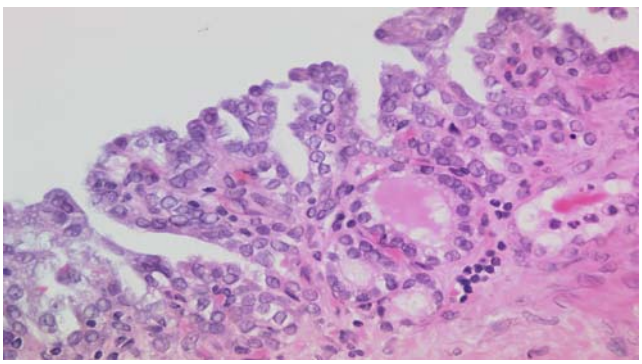

Fig. 1 Haematoxylin and eosin (H & E) section of struma ovarii showing nuclear grooving and ground glass appearance

Thyroid ultrasound was normal.

She underwent laparoscopic left salpingo-oophorectomy and omental and peritoneal biopsies. The pelvis looked normal with no macroscopic evidence of any disease in the peritoneal cavity except a few postoperative filmy adhesions. Histology of the peritoneal biopsies, ovary and the fallopian tube did not show any evidence of residual struma ovarii and thyroid tumour.

Total thyroidectomy was performed and the thyroid tissue did not show any evidence of malignancy.

She was seen in the gynaecology oncology outpatients as follow-up. She had made a good postoperative recovery and was on thyroxine replacement therapy.

She was later admitted to the Oncology and Radiology Department and received 3,000-Mbq dose of radio iodine. Further follow-up management plan was to subject her to radioiodine challenge scanning and check her thyroglobulin levels.

She remains disease free 20 months after surgery and is still being followed up closely with clinical, radiologic and laboratory examinations.

Discussion

Malignant transformation is an extremely rare complication of struma ovarii, occurring in 5–10% of cases. Several different types of thyroid carcinoma arising in struma ovarii have been described, of which the papillary variety is the most common. It mostly occurs in the fifth decade of life and is most common on the left side [1]. It is important to eliminate thyroid as the primary site of neoplasia.

Clinical presentation is nonspecific with findings similar to other ovarian tumours and the diagnosis is usually made postoperatively. Struma ovarii have been infrequently associated with thyrotoxicosis, and only 5% of cases have evidence of thyroid overactivity [2]. It has also been associated with pseudo-Meig's syndrome [2] which is comprised of ascites and hydrothorax, and the case under discussion was reported to have free fluid in abdominal cavity on ultrasound scan; however, there was no clinical or radiological evidence of hydrothorax. Ovarian tumour markers in our case were not done as there was low clinical suspicion and was dealt by the surgical team initially. Increased levels of Ca125 have been reported in struma ovarii. [3]

Laparoscopy in our case was performed under the care of general surgeons as the standard practice in our surgical unit is assessment by laparoscopy and, if appropriate, treatment by the same. The appearances of the cyst at laparoscopy were benign, namely, thin wall, no septations or solid component, and so frozen section was not obtained. Laparoscopic management was deemed appropriate as the

ovarian cyst had no macroscopic features to suggest malignancy and had already ruptured with gross spillage.

The histologic criteria for malignancy in struma ovarii are controversial. The presence of malignant nuclear features designated as ground glass overlapping nuclei and nuclear grooves has been accepted as the most crucial criteria for diagnosis of papillary carcinoma [1]. In our case, the microscopic features were typical of papillary carcinoma and immunoperoxidase staining was positive for thyroglobulin and TTF-1.

Due to rarity of malignant struma ovarii, there is no consensus to the extent of surgical management. Currently, unilateral salpingo-oophorectomy is the minimum surgery that should be considered [3]. The index case had only left salpingo-oophorectomy on confirmation of diagnosis of papillary carcinoma of struma ovarii as there was no evidence of any macroscopic disease of the other ovary and the woman wished to preserve her fertility. More extensive surgery like bilateral salpingo-oophorectomy and total abdominal hysterectomy with or without omentectomy and lymph node dissection can be performed depending on the extent of the disease and the fertility retaining wishes of the patient [3, 4].

There is still a controversy regarding the aggressiveness of further management. Additional interventions that may be considered are thyroidectomy with or without iodine-131. In our case, total thyroidectomy was performed followed by radioiodine treatment. Thyroidectomy offered the following advantages: it helped to exclude primary thyroid carcinoma, facilitated whole-body radioiodine scan and will help long-term monitoring by thyroglobulin estimation. Radioactive iodine treatment following thyroidectomy ablates microscopic foci of malignancy if any and also residual thyroid tissue, thus improving follow-up with thyroglobulin estimation, and hence the index case received 3,000 Mbq radioiodine.

Metastasis occurs only in 5–6% [3], mainly to the peritoneum, liver, lung, bone, spleen and the contralateral ovary [2]. Hence, long-term follow-up is required. Our case remains disease free 20 months after treatment; however, she is still under close follow-up.

New methods like thyroglobulin mRNA could prove valuable in those patients who develop antithyroglobulin antibodies [3].

To conclude, despite the rarity of the condition, adequate management can be achieved by combining surgical treatment and further interventions currently used for the management of primary thyroid carcinoma. Long-term outcome for papillary variant seems to be more favourable than its follicular counterpart.

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