A rare case of papillary thyroid carcinoma arising in struma ovarii

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Introduction

Mature cystic teratomas account for 15 to 20% of them[1-4]. Thyroid tissues are found in 15 to 20 percent of those[2,3] but the term "struma overii" where the major proportion of the tumour comprised of thyroid tissue accounts for only 3 to 5 percent[5]. These are usually benign but rarely show malignant transformation (Malignant Struma Ovarii)[5]. There are no protocols or guidelines for the management of these tumours due to its rarity, and management should be individualized. This is a case of papillary carcinoma of thyroid arising in a struma ovarii which was managed by conservative surgery. We are presenting this, due to its rarity.

Case history

A 30 year old mother of one child presented with urinary symptoms and found to have a pelvic mass. She had no other co-morbidities and not had a family history of gynaecological, colonic or thyroid malignancies. She was found to be clinically euthyroid. On examination she was found to have an adenexel mass with normal size uterus. Her thyroid gland was not enlarged and respiratory system clinically normal with no evidence of pleural effusion.

Ultrasound scan of the abdomen and pelvis revealed a large bilocular cyst in left adenexiae occupying the pouch of Douglus. Right ovary and uterus appeared normal and left ovary not visualized. Rest of the abdomen was normal with normal kidneys and liver. No free fluid noted.

Since she was euthyroid initial thyroid functions were not done.

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She underwent a laparotomy and during surgery bilateral ovarian cysts noted, right mass measuring 10 cm×8 cm and left mass measuring 8 cm×6 cm. Their appearance was suggestive of dermoid cysts. Rest of the abdomen and pelvis was normal including a normal size uterus. There was no free fluid. Due to the benign nature and reproductive wishes bilateral cystectomy and reconstruction of both ovaries done. The pathological examination showed 3 cysts, measuring 90 mm×75 mm×60 mm, 50 mm×45 mm×40 mm and 40 mm×30 mm×30 mmin in size with outer smooth walls. The first and the third cysts contained hair and sebaceous materials, while the second specimen contained friable solid tissue.

The first and the third cysts turned out to be mature cystic teratomas showing squamous epithelium, sebaceous glands, hair follicles, respiratory type epithelium and cartilage on microscopic examination. They did not contain immature elements. The second cyst showed a papillary carcinoma composed of haphazardly arranged papillae lined by cells showing typical nuclear features of papillary carcinoma of the thyroid indicating papillary thyroid carcinoma arising in a struma ovarii. There was no vascular invasion.

Thyroid function tests were performed post operatively and became normal (Free triiodothyronine 2.44pg/ml (1.5-4.1), Free thyroxine 1.87ng/dl (0.8-1.9) and TSH level 0.877miu/ml (0.4-4.0). Thyroglobulin level 0.8ng/ml (0.83-68).

Ultrasound scan of the thyroid gland only revealed a small heterogeneously hypoechoic small lesion on the left lobe, of which FNAC revealed benign colloid nodule.

Further post operative intravenous contrast CT scan of the abdomen and pelvis revealed normal size uterus and prominent ovaries, showing heterogeneous attenuation pattern and heterogeneous enhancement pattern following intravenous contrast. But there were no focal mass lesions in the ovaries and no lymphadenopathy detected.

Iodine^{131} whole body scan revealed nodular tracer uptake in the region of thyroid gland only.

She was followed up with 3 monthly USS, and one year later with a whole body PET scan, and all found to be normal.

Discussion

Ovarian teratomas accounts for up to 20% of ovarian tumours (1-4). Up to 15% to 20% of teratomas contains thyroid tissue[2,3]. But struma ovarii, which refers to a teratoma in which thyroid tissue is the, predominant or the sole component of the tumour, or forms the grossly recognizable component of a more complex teratoma or at least making up more than 50% of tumour, is very rare, accounting only 3 to 5% of teratomas[2,3].

Teratomas can undergo malignant transformation rarely, and commonest germ cell line that undergo malignant transformation is squamous cell carcinoma. Histological features of thyroid carcinoma is found in struma ovarii in about 5%, known as malignant struma ovarii[2,3]. The routine microscopic nuclear features of thyroid carcinoma are used for the diagnosis. Immunohystochemical staining is also can be used for the confirmation of the diagnosis as well. The diagnosis is almost always by histology following laparotomy which was done for ovarian tumour. Even with facilities for frozen section, it will only reveal the presence of thyroid tissue in teratoma and therefore definitive diagnosis is almost always post operative.

Histological criteria for the diagnosis of malignant struma ovarii are based on the guidelines for primary papillary thyroid carcinoma. There are three histological types. The most common reported type is papillary carcinoma which accounts for 44% of malignant struma ovarii. The features of these tumors include optically clear or ground-glass overlapping nuclei with nuclear grooves. The second type is follicular carcinoma which accounts for 30%, which is diagnosed when cells containing mitotic figures begin forming follicles and capsular or vascular invasion is noted .Third type is follicular variant of papillary carcinoma, which accounts for 26% of malignant struma ovary[1,3,6,] which might have reported as follicular carcinoma in early case reports[7].

Metastasis of the malignant stuma ovarii is rare, and even rarer with those with no vascular invasion and capsular breach. Its metastasis is like other malignant ovarian tumours, and The tumour can spread directly to the omentum, peritoneal cavity, and contra lateral ovary. It can also metastasize to the pelvic or para-aortic lymph nodes via lymphatics or hematogenously to the bones, lungs, mediastinum, liver or brain[8,9].The patient can be evaluated by performing a Iodine 131 nucler medicine scan for occult secondarys and the sensitivity of which is improved by performing a thyroidectomy, and by imaging techniques[5].

Presence of malignant thyroid tissue in ovary should also raise the suspicion of primary thyroid malignancy, giving rise to ovarian secondary. But it is unlikely to present in a teratoma.

There are no definite treatment protocols for the management of struma ovarii due to the rarity of the disease. Surgical treatment may include maximum cytoreductive surgery, including total abdominal hysterectomy, bilateral salpingo oophorectomy, omentectomy, lymph node dissection and peritoneal fluid washings for cytology followed by adjuvant therapy, or more conservative surgery by means of unilateral salpingo oophorectomy or cystectomy, depending on the clinical presentation, age of the patient and the reproductive whishes of the patient.

How ever there are no good evidence to support which method is best. Considering rarity of metastatic disease, many clinicians perform conservative surgery, and adjuvant therapy is preserved for those patients with locally advanced, recurrent or metastatic disease[9,10].

Reported adjuvant therapy includes chemotherapy and thyroidectomy followed by ablation of residual tumours and metastatic deposits using radio active Iodine. Performing thyroidectomy not only exclude thyroid malignancy but also make the treatment more effective as well.

Long term follow up is needed for them especially in those who had conservative surgery. It can be carried out initially by excluding possible secondaries, during surgery and imaging techniques CT scan and Iodine^{131} nuclear medicine scanning, followed by serum thyroglobulin levels and If any evidence of elevated thyroglobulin to perform nucler medicine imaging. Recurrence of tumour on average is reported after 4 years of surgery and overall, the survival rate for all patients was 89% at 10 years and 84% at 25 years, indicating the need for routine long-term follow-up11. Therefore some authors recommend follow up, up to 10 years with thyroglobulin measurements and imaging techniques[3,11].

Our follow up plan is to perform USS to assess ovaries and pelvic masses every 3 months, thyroglobulin levels annually for 10 years. After one year, her PET scan and Contrast CT scan revealed no

secondaries and her thyroglobulin level found to be normal. During two years of her post operative period she remained asymptomatic and disease free, and planning for a pregnancy.

Conclusion

struma ovarii is a rare type of monodermal teratoma, where malignant transformation is even rarer. There are no management protocols available for the management due to its rarity and only evidence available are from case reports and few case series. There fore its management should be individualized and follow up plans should be drawn to monitor the occurrence of recurrences and metastasis.

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