Malignant Struma Ovarii: Case Report and a Review of the Literature.

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Abstract
Struma ovarii is a rare tumor. Malignant change in this tumor is even rarer. While clinical presentation is protean, a preoperative diagnosis is equally difficult. Diagnosis is confirmed by histology. Treatment of malignant struma ovarii is by surgical resection of the tumor or total abdominal hysterectomy and bilateral salpingo-oophorectomy. A total thyroidectomy allows radioactive iodine ablation and the use of thyroglobulin as marker to monitor recurrence. However due to rarity of the disease, standard treatment is still controversial. We present a case of malignant struma ovarii, followed up for five years after treatment, and a review of the literature on the disease. The patient was treated with surgery and radiation. She is still free of disease after 5 years.

Key Words: Malignant Struma Ovarii, Ovarian Teratoma, Papillary Thyroid Carcinoma

Introduction
Struma ovarii is a rare tumor, accounting for approximately 1% of all ovarian tumors. It is a monodermal variant of ovarian teratomas in which thyroid tissue is the major constituent (>50%). Struma ovarii constitutes only 2.7% of mature cystic teratomas. Malignant struma ovarii is even much rarer, being seen in just about or less than 5% of struma ovarii.

Clinical Case
We present a 44-year-old Para 2+ female who was first seen at an Indian hospital about June 2005 when she presented with a history of worsening lower abdominal pains. The pain was not relieved by analgesics. There was no known precipitating or aggravating factor. There was no unusual vaginal bleeding or discharge. Her last confinement was 26 years prior to presentation. There was no past or current use of intrauterine contraceptive devices. Her menstrual cycle had been fairly regular. She was not a known peptic ulcer patient.

Ultrasonography of the abdomen showed a left adnexal mass measuring 37mm x 35mm in the transverse plane. The mass was anechoic and had posterior acoustic enhancement. The uterus and contralateral ovary were essentially normal but minimal fluid was seen in the pouch of Douglas. An impression of a functional left ovarian cyst was made with a request for clinical correlation.

Progression of patient’s symptoms led to a laparotomy. Operative findings showed normal sized right ovary with small cysts which contained serous fluid. The left ovary was enlarged, had multicellular cysts containing sebaceous materials, hair and serous fluid. The uterus was grossly unremarkable.

A total abdominal hysterectomy (TAH) with bilateral salpingo-oophorectomy (BSO) was carried out. The histopathology revealed that the left ovary had features of mature cystic teratoma with malignant transformation in thyroid tissue consistent with papillary thyroid carcinoma. Post operatively, serum alpha fetoprotein level was 7ng/ml, CA-125 was 2U/ml, â HCG was <0.05mIU/ml. CT scan of the abdomen scan as well as thyroid ultrasound scan done two months post operatively were normal. A base line thyroid function test showed T3 (1.7µg/dl), T4 (9.5µg/dl), TSH (5.3µIU/ml). This was followed by a pertechnetate (TcO4) thyroid uptake test, which showed uniform uptake in both thyroid lobes. No abnormal extra-thyroidal uptake was seen. A near total thyroidectomy (NTT) was

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abnormal vaginal bleeding, ascites, hydrothorax our patient where typical features papillary pains, palpable lower abdominal mass, 16). This however seemed not to be the case in presence of struma ovarii are lower abdominal may rely on the presence of cellular atypia (15, 16) even though it is a Devany (13) and co-workers have urged the use of this marker obviates the need for the diagnostic scan to confirm ablation, we did not have cause to repeat a 131 WBS in the 5 years of following up this patient. The thyroid function tests, serum thyroglobulin and anti thyroglobulin antibody (ATA) assays have so far remained excellent indices to monitor recurrence.

The widely accepted tumor marker of ovarian cancers CA-125 is found to be increased in 80% of epithelial ovarian carcinomas. This marker is also elevated in other tumorous lesions of the endometrium, intestines, breasts, and lungs as well as in non malignant related gynecologic conditions, thus indicating that there is a limit to the clinical application of CA-125 as a tumor marker of ovarian neoplasms. Moreover, it has been postulated that increased levels of CA-125 is not a direct consequence of the presence of malignant tumor, but rather a secondary effect due the presence of ascites. (23)

Management of patients who desire fertility might be challenging. (9) Another challenge sometimes encountered fact is patient’s refusal to accept thyroidectomy, as shown by Yücesoy et al (20). Patients treated without thyroidectomy with survival rates of 12-180 months have been reported. (21) In struma ovarii, peritoneal fluid levels of thyroglobulin might indicate an intra-abdominal extension or recurrence. Follow up by means of this marker obviates the need for repeated 131 scanning and the unpleasant hypothyroid symptoms inherent to the interruption of thyroid substitution. (22) Except for the diagnostic scan to confirm ablation, we did not have cause to repeat a 131 WBS in the 5 years of following up this patient. The thyroid function tests, serum thyroglobulin and anti thyroglobulin antibody (ATA) assays have so far remained excellent indices to monitor recurrence.

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In the case series by Yoo et al (12), it was observed that CA-125 was increased in 4 among 13 patients with struma ovarii (3 malignant and 10 benign). This comprised of 1 out of the 3 malignant cases and 3 of the 10 benign cases. The elevation in the malignant case was minimal. This, coupled with the presence of elevations in benign struma ovarii cases caused them to propose that serum CA-125 measurements are of little clinical value in struma ovarii patients. In the case we report the patient had no ascites and the CA-125 level was normal.

Table 1: Comparative Findings in the Literature* versus those seen in the Patient.

<table>
<thead>
<tr>
<th>Features*</th>
<th>Index Patient</th>
<th>Features*</th>
<th>Index Patient</th>
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</thead>
<tbody>
<tr>
<td>Age distribution in years (n=34)</td>
<td>21-80</td>
<td>44 years</td>
<td></td>
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<tr>
<td>Patients aged 41-50 (n=34)</td>
<td>26.6%</td>
<td>Yes</td>
<td></td>
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<tr>
<td>Palpable abdominal mass (n=34)</td>
<td>23.5%</td>
<td>No</td>
<td></td>
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<tr>
<td>Abdominal Pain (n=34)</td>
<td>20.6%</td>
<td>Yes</td>
<td></td>
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<tr>
<td>Vaginal Bleeding (n=34)</td>
<td>8.8%</td>
<td>Yes</td>
<td></td>
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<tr>
<td>Increased CA125 (n=13)</td>
<td>30.8%</td>
<td>No</td>
<td></td>
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<tr>
<td>Benign cyst at US scan (n=34)</td>
<td>8.8%</td>
<td>Yes</td>
<td></td>
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</table>

*Yoo et al (12)

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References


