

High dose Radioiodine Therapy Preceded by Fertility Preservation Surgery in Metastatic Malignant Struma Ovarii: A Tale of Endurance and Prudent Management

Abstract

Struma ovarii is a type of mature ovarian teratoma which accounts for roughly 0.5%–1% (1) of all ovarian tumours and approximately 3% of all ovarian teratomas (2). To be classified as struma ovarii, more than 50% of the tumour must be comprised of thyroid tissue (3). Malignant struma ovarii being rare, no proper guidelines exists regarding its surgical approach or postoperative management. Metastatic malignant struma ovarii, in addition to radical surgery for ovarian mass will require total thyroidectomy to facilitate high dose radioiodine therapy. Here we present the case of a newly married, nulliparous, young lady in her third decade who was diagnosed with malignant struma ovarii with metastatic deposits in fallopian tube and extensive deposits in mesentery and peritoneum. She underwent cryopreservation of embryos followed by bilateral salpingo-oophorectomy + omentectomy + stripping of peritoneum over bladder, abdominal side walls, pelvic peritoneum + appendectomy with preservation of uterus. Total thyroidectomy was done simultaneously. Subsequently she underwent high dose radioiodine therapy. Complete ablation of the residual metastatic deposits were achieved by one sitting of therapy.

Keywords: Fertility preservation, metastatic malignant struma ovarii, radioiodine therapy

Introduction

Struma ovarii is a rare type of mature ovarian teratoma where significant portion of it is comprised of thyroid tissue. It comprises roughly 0.5%–1%^[1] of all ovarian tumours and approximately 3% of all ovarian teratomas.^[2] Though thyroid tissue can be seen in 5%–15% of dermoid tumours, teratoma to be classified as struma ovarii, more than 50% of the tumour must be comprised of thyroid tissue.^[3] Peak age of incidence is in fifth decade and presenting symptoms are often nonspecific. Many patients remain asymptomatic for a considerable time period leading to delay in diagnosis. Patients may present with abdominal pain, hyperthyroidism, menstrual irregularities, abdominal mass or ascites. Management of benign struma ovarii is surgical removal of ovarian cyst or mass. In euthyroid patients, diagnosis is often made post operatively in the histopathology of resected specimen. Literature on the incidence of malignancy in struma ovarii is limited due to the rare nature of this condition.^[4] When diagnosis is made

post operatively in ovarian cystectomy specimen, further radical surgery with Total abdominal hysterectomy + bilateral salpingo oophorectomy + omentectomy along with Total thyroidectomy may be required in cases where histopathology shows differentiated thyroid carcinoma in the resected specimen.^[5] We present the case of a married, nulliparous lady, who was diagnosed with Struma ovarii and multiple peritoneal metastatic deposits from highly differentiated follicular carcinoma.

Case Report

A 29-year-old, nulliparous lady, presented with history of abdominal pain, localised to right iliac fossa. History of laparoscopic ovarian cystectomy 10 years ago where histopathology was reported as endometriotic cyst. Ultrasound showed bilateral complex adnexal masses. Based on the prior history, an imaging differential of atypical endometriosis with endometrioid ovarian neoplasm was considered.

Contrast enhanced magnetic resonance imaging (MRI) [Figure 1] revealed no focal

Shagos Gopalannair Santhamma, Valam Puthussery Vipin¹, Jem Kalathil², Nita Mary John³

Departments of Nuclear Medicine and PETCT, ¹Endocrinology, ²Surgical Oncology and ³Clinical Pathology, Aster Medcity, Kochi, Kerala, India

Address for correspondence:

Dr. Shagos Gopalannair Santhamma, Department of Nuclear Medicine and PETCT, Aster Medcity, Kochi - 682 027, Kerala, India. E-mail: shagosgs@gmail.com

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myometrial lesions. Multiple solid cystic bilateral adnexal masses with few solid enhancing nodules within the cysts visualised. Normal ovaries not separately identified. Majority of the cysts had irregular thick septae. Solid nodules were isointense in T2 signal with low ADC signal with moderate post contrast enhancement. Loss of fat planes of adnexal masses with lateral margins of uterine body noted. Multiple enhancing peritoneal implants in the pouch of Douglas (POD), bilateral iliac fossae and perivesical spaces also seen Figure 1.

CA 125 was marginally raised 58.48 U/mL (<35.00 ECLIA). She underwent open bilateral ovarian cystectomy. Histopathology [Figure 2] showed Struma ovarii with multiple peritoneal deposits; consistent with highly differentiated follicular carcinoma. Immunohistochemistry [Figure 3]: Cells were positive

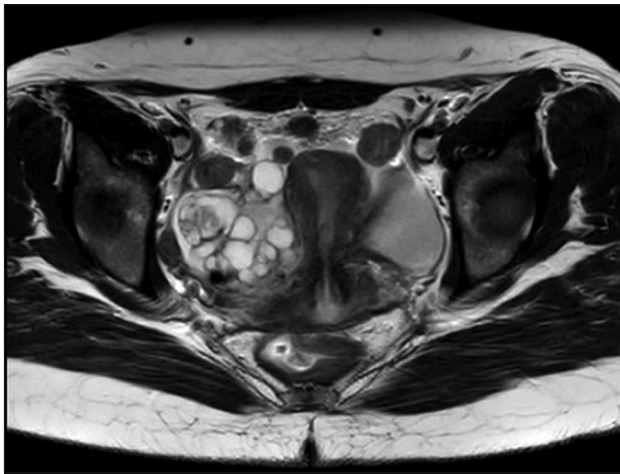


Figure 1: Contrast enhanced Magnetic resonance imaging with T2W axial images showing normal size, shape and signal intensity of uterus with no focal myometrial lesions. Bilateral solid cystic adnexal masses with few solid enhancing nodules within the cysts noted with normal ovaries not visualised separately. Right solid cystic mass measured 7.6 cm x 3.6 cm x 6.3cm and left solid cystic adnexal mass measured 5.0 cm x 3.8 cm x 5.8cm

for thyroid transcription factor-1 (nuclear) and thyroglobulin (cytoplasmic). Ultrasonography neck revealed normal thyroid gland Figures 2 and 3.

Thyroid function tests showed subclinical hyperthyroidism. After multi disciplinary tumour board discussion, decision of staging laparotomy and total thyroidectomy with a view of treating her with adjuvant high dose radioiodine therapy was taken. As she desired fertility preservation, follicle aspiration and cryo preservation of 2 embryos were done prior to surgery. Subsequently bilateral salphingo-oophorectomy + omentectomy + stripping of peritoneum over bladder, abdominal side walls, pelvic peritoneum + appendectomy with preservation of uterus carried out. Histopathological reports: Struma ovarii (bilateral) with extensive extra ovarian metastasis; consistent with Highly differentiated Follicular carcinoma, involving left fallopian tube, omentum, splenic flexure,

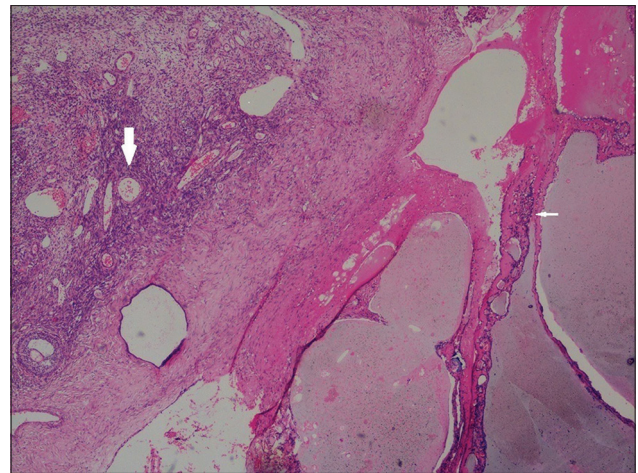


Figure 2: Histopathology Section (Haematoxylin and Eosin stain) showing ovarian tissue on the upper left hand corner (bold arrow) and colloid filled follicles of varying sizes lined by fairly uniform thyroid follicular cells on the lower right hand corner (thin arrow)

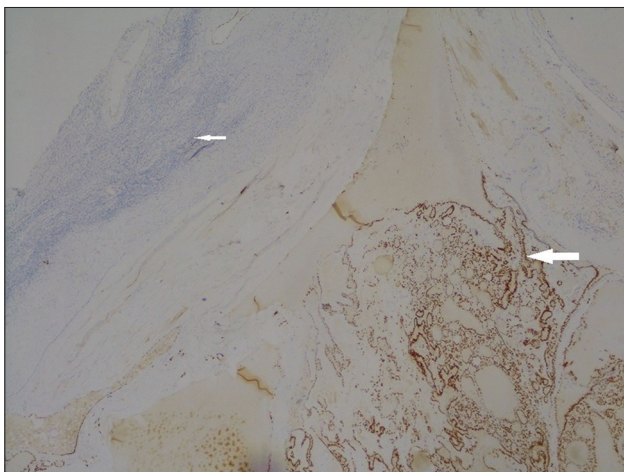


Figure 3: Immunohistochemistry for Thyroid transcription factor-1 showing nuclear positivity in thyroid follicular cells (bold arrow). Ovarian tissue at the upper left hand corner is negative (Thin arrow)

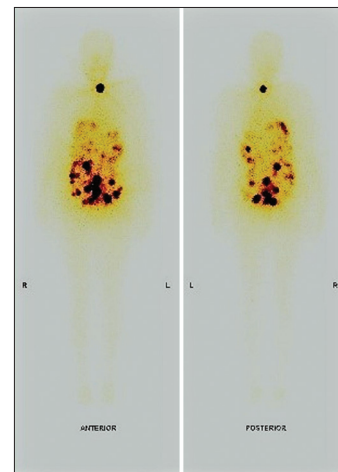


Figure 4: Post therapy whole body I 131 scan showing I131 tracer uptake in the residual thyroid tissue and multiple (serosal) deposits in the intestinal loops in abdomen

sigmoid colon deposit, mesentery, small bowel deposit, right and left lateral peritoneum, right and left pelvic (POD) peritoneum, mesoappendix and bladder peritoneum. Total thyroidectomy specimen showed no significant pathology.

Pre therapy Iodine-131 scan showed residual thyroid tissue with multiple small I-131 concentrating deposits on the serosal surface of bowel. Stimulated serum thyroglobulin value was 70.51 ng/ml, antithyroglobulin Ab 27.3 IU/ml and thyroid stimulating hormone (TSH) more than 100 μ IU/ml.

Figure 4 High dose I-131 therapy was carried out with 6475 MBq of I-131 orally. Post therapy scan [Figure 4] showed I-131 uptake in all the metastatic sites as seen in pretherapy scan.

Follow up I-131 scan [Figure 5] after 6 months showed complete resolution of I-131 concentrating serosal deposits with no new metastatic foci. Follow up stimulated serum Thyroglobulin was 0.71 ng/ml and Serum TSH more than 100 μ IU/ml Figure 5.

Discussion

Struma ovarii being a rare ovarian tumour with atypical presentation, there is a paucity of data in published literature regarding surgical management, adjuvant therapy and follow up evaluation. In the absence of thyrotoxicosis, the diagnosis is often delayed until patient presents with symptoms of ovarian torsion or ascites. CA-125 is of little value in differentiating struma ovarii from other ovarian epithelial tumours as increased CA-125 levels can be seen in struma patients due to secondary effects of ascites. Unless a proper radiological investigation with preoperative MRI is carried out, patients with benign struma ovarii may be subjected to unwarranted radical surgery. In young females with malignant struma ovarii, if fertility preservation is preferred, conservative surgery needs to be adopted with unilateral salpingo-oophorectomy or ovarian cystectomy based on the preoperative imaging findings.

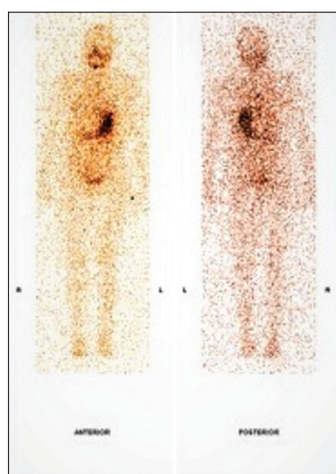


Figure 5: Follow up whole body I 131 scan showing complete ablation of residual thyroid tissue with no abnormal focus of I31 tracer uptake in the abdomen

No proper guidelines exist regarding postoperative management of malignant struma ovarii with histopathology showing differentiated papillary or follicular features similar to thyroid malignancy. Total thyroidectomy even in the absence of radiological evidence of thyroid malignancy is mandatory in metastatic malignant struma ovarii to facilitate adjuvant radioiodine therapy. In the absence of metastatic disease role of total thyroidectomy is debatable.

Adjuvant high dose I-131 therapy for metastatic malignant struma ovarii has been reported^[5] with excellent therapeutic response. Metastasis is commonly intraabdominal (5%–23%) in the omentum or peritoneum, but has been reported to liver, lung, brain and bones through blood stream spread.^[6] Follicular carcinoma's have more propensity for blood stream spread when compared to papillary subtype.^[7] Ablative doses of radioiodine is decided based on I-131 scan findings and stimulated serum thyroglobulin levels. Radioiodine therapy facilitates detection of disease recurrence by doing follow up iodine scan and stimulated thyroglobulin values.

In our patient total thyroidectomy facilitated adjuvant high dose radioiodine therapy for the metastatic disease. Normalisation of whole body Iodine-131 scan after single sitting of high dose Iodine-131 therapy with fall in stimulated serum thyroglobulin value to <1 ng/ml confirmed successful ablation. The cryopreserved embryos were implanted a year later using assisted reproductive techniques and a healthy baby was delivered by elective caesarean section at 36 weeks.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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