Case Report

Struma-ovarii – Literature Review and A Case Report of Malignant Struma-ovarii

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Struma-ovarii are specialized teratomas of thyroid tissue. We report here a case of papillary thyroid carcinoma in struma-ovarii in a morbidly obese woman with review literatures. The ultrasonography revealed a large complex multiloculated right ovarian cyst 10.6x11.1 cm. with solid components. The tumour was removed by laparoscopic salpingo-oophorectomy. On macroscopy, tumour was multiloculated cyst 15x15cm, filled with gelatinous material without papillary excretion and intact capsule. The histological sections demonstrated follicular pattern of papillary thyroid carcinoma's characteristic optically clear nuclei with thickened nuclear membrane, grooving and cellular pleomorphism. The final diagnosis was malignant struma-ovarii, FIGO- Stage-IA. The recovery was uneventful, remaining disease-free for over years. The prognosis is good after conservative surgery Oophorectomy in malignant struma-ovarii when limited to the ovary and the capsule is intact.

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Struma-ovarii is a cystic teratoma of the ovarii[1]. The term Struma-ovarii is assigned to such teratoma of the ovary where thyroid tissue is predominant or its sole consistituent[2].

Gottschalk S were the first to publish a case of true struma-ovarii in late 1890s and considered it arising from ovarian follicles[3]. Gottschalk also noted malignant area in the teratoma and designated it as Folliculoma Malignum. Struma-ovarii occurs at any age between 18-84 years, the peak incidence is in the fifth decade of life, it is more common in left ovary.

There is no biomarker for the Struma-ovarii. Malignancy in struma-ovarii is rare. Lack of universally accepted criteria for malignancy and the varied clinical course contribute to discrepancies in reported cases of malignant struma-ovarii[4].

The actual diagnosis of stuma-ovarii is rarely made until tumour section is examined under microscope. Histopathology of the resected tumour permits diagnosis of struma-ovarii and at the same time confirm or exclude malignancy[1].

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Editor's Comment :

- Struma Ovarii are rare specialized teratoma thyroid tissue. They can undergo malignant change 5-10%.
- It does not have definite clinical or imaging characteristics that differentiates it from other ovarian tumours.
- Serum LDH and serum thyroglobulin level may be used to screen it.
- Conservative surgery Salpingo-Oophorectomy can be offered when tumour is limited to one ovary with intact capsule in young patient with good prognosis.

Case Report

A 34-year-old patient presented with complaint of lower abdominal pain for 6 months duration and irregular menstrual bleeding for 4 months. She was morbidly obese with a body mass index (BMI) of 46 kg/m^2. She was on oral contraceptives and had previous abortion and a Lower Uterine Segment Caesarean Section. She was presented with a large abdominopelvic lump of size of 22 weeks pregnancy. Ultrasonography (USG) revealed presence a large complex multiloculated cyst (10.6 x 11.1 cm) in size with septations and solid components (Fig 1-A). Left ovary and the uterus were normal in size.

Triphasic computerized extracorporeal tomography (CECT) (Fig 1-B) confirmed USG findings of multiloculated cyst in the right ovary showing internal density ranging from 33 to 63 HU with solid portion and hemorrhagic fat density, the diagnosis was mature cystic teratoma of right ovary. Cancer Antigen (CA) 125 was within normal range of 6.34 U/mL. The tests of urine, blood, thyroid function, PAP smear from cervix were all normal. Fasting blood sugar was high (155 mg/dl), Serum Lactate Dehydrogenase (502 U/L) & Thyroglobulin level (32.7 ng/Ml.) were normal.

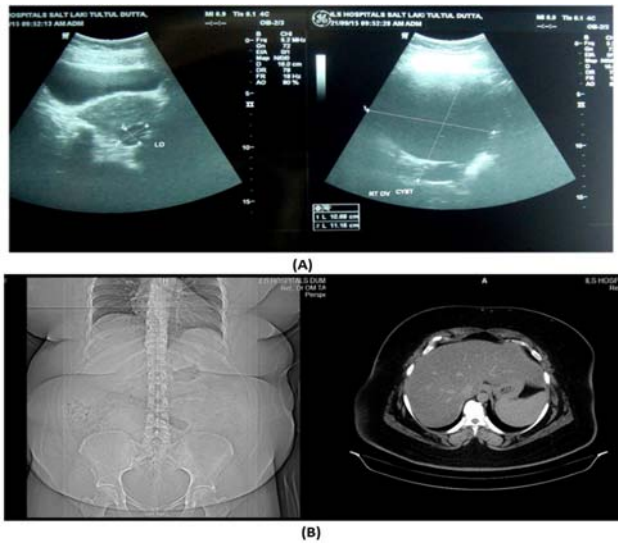

Fig 1 — (A) USG: Large complex multiloculated right ovarian cyst with septations and solid components. (B) – CT scan of lower abdomen: The large multiloculated cystic space occupying lesion (SOL) in the right adnexa showing internal density.

Laparoscopy revealed a large cyst in right ovary, left ovary was normal.

The tumour was removed by Right Salpingo-Oophorectomy by laparoscopic surgery.

On macroscopy, the tumour was a multiloculated cyst 15 x 15 cm. in size filled with gelatinous material (Fig 2-A). There was no papillary excretion or no break in the capsule. The histology demonstrated follicular pattern of papillary thyroid carcinoma characteristic optically clear (ground glass) nuclei with thickened membrane and grooving and cellular pleomorphism (Fig 2-B).

The final diagnosis made was Malignant struma-ovarii, stage – IA, Federation of International of Gynaecologists and Obstetricians (FIGO).

The patient made uneventful recovery and went home on the fourth day of operation. Clinical findings, USG of Neck, CECT Abdomen, Chest X-Ray, Thyroid profile and Thyroglobulin level all were normal at the time of discharge.

Discussion

Thyroid tissue within an ovarian dermoid was first described by Bottlin in 1889 and Pick in 1901 postulated such dermoids as teratomas[4]. Meyer in 1903 first coined the term "Struma-ovarii Colloids". Pick[5] believes that in struma-ovarii the thyroid tissue proliferates while other elements are suppressed in such teratoma.

Histologically struma-ovarii can resemble thryroid adenoma follicular or embryonal type or thyroid carcinoma. Malignancy should be suspected when there is ascites and CA 125 is elevated, Presentation of the struma-ovarii is like that of any other teratoma of ovary and is usually benign. Malignancy in it is reported as 5-10%. Only 21 malignant struma-ovarii with metastasis have so far been reported.

K Zied *et al*[6] reported three cases of struma-ovarii with different presentations.

One a 19 years old girl had menstrual irregularity and a large cystic pelvic mass.

The other was 31 years old, came for investigations of infertility. Routine USG revealed a small cyst in the right ovary and Magnetic Resonance Imaging diagnosed it as teratoma. Third one was 45 years old, parous had intractable menorrhagia and no pelvic mass. She was treated by Hysterectomy with bilateral salpingo-oophorectomy. The right ovary showed, multiple small cysts filled with brown gelatinous material which on microscopy showed evidence of struma-ovarii. Thus struma-ovarii does not have definite clinical or imaging characteritics that differentiates it from other ovarian tumours.

Malignant transformation in Struma-ovarii is extremely low[7]. Hard fixed nodule in the pouch of douglas is not usual neither is ascites. Ascites is not present though the struma-ovarii is malignant because the tumour capsule is not broken down and there is no spread malignant deposits in the general peritoneal cavity. Further ascites could be present in absence of cancerous transformation of the struma where presentation of the case is of pseudo meig's syndrome marked.

Marcus *et al*[8] in their series of seven cases of struma-ovarii described three main histological groups namely: Thyroid tissue predominant in a benign cystic teratoma, the thyroid tissue in cystic adenoma and a pure struma-ovarii.

There is difference in opinion as to what microscopic appearance of the tissue constitute cancer in struma-

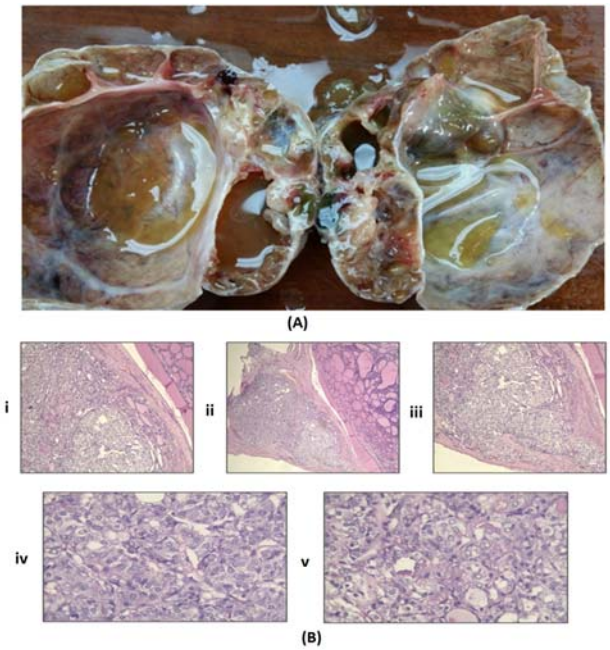

Fig 2 — (A) Cut section of the cyst: showed multiloculated cyst filled with greenish yellow gelatinous material. (B) – Histopathology: Histopathological examination illustrating poorly differentiated thyroid carcinoma arising in struma-ovarii (i) Focal papillary architecture; (ii) Foci of follicular variant of papillary carcinoma in struma-ovarii; (iii) Follicular arrangement; (iv) Nuclei showing grooving; (v) Optically clear nuclei with thickened nuclear membrane.

ovarii. Smith GF[9] held that blood vessel invasion is the only definite criteria of malignancy. Vessel invasion though is difficult.

Vigorous criteria for the diagnosis of malignancy include confirmation of capsular invasion and or metastasis. But evaluation of capsular invasion in struma-ovarii is difficult and can not be used as an essential criteria of malignancy[4].

In general malignancy is diagnosed on the basis of sufficient degree of cellular pleomorphism cytological atypia and mitotic activity. Zakhem *et al*[1] describes two cases of malignancy in struma-ovarii based on histological and nuclear alterations. In his series Devaney[2] describes 14 cases of struma-ovarii using histological criteria of mitosis and ground glass nuclei.

Possibility of metastatic thyroid carcinoma was eliminated in the present case from history, clinical examination, USG of thyroid gland and thyroid functional profile and Thyroglobulin level.

Metastatic struma-ovarii from thyroid carcinoma is rare but from malignancy has to be excluded arising in struma-ovarii. Logani *et al*[10] described a woman who manifested of metastatic papillary thyroid carcinoma in the ovary. The woman was 34 year old and underwent total thyroidectomy for papillary carcinoma of the thyroid gland. Serum thyroglobulin showed significant increase in level from 1.6 ng/ml. to 3.4 ng/ml. in about 3 years period. On follow up of the case, radio imaging with I-131 showed its concentration in the neck and pelvis. Magnetic Resonance Imaging revealed a cystic mass 6 x 4 cm. of size in the left adnexa. The mass removed by Hysterectomy. Histopathology showed it struma-ovarii with features of papillary thyroid carcinoma.

All pathological pattern of malignancy are found in struma-ovarii, but papillary follicular carcinoma is not common. Navarro *et al*[4] in the review of malignant struma-ovarii identified 16 papillary carcinomas, 14 follicular carcinomas and 5 combined papillary carcinoma. To this list they added one of their own case of struma-ovarii which is papillary variant of follicular carcinoma. The case of malignant strum ovarii presented here likewise is the papillary variant follicular carcinoma. Invasion of capsule could not be demonstrated but diagnosis of malignant struma-ovarii is reasonable from the presence of cellular pleo morphism and characteristic nuclear pattern and so may be included in the list of papillary variant of follicular carcinoma in struma-ovarii.

Prognosis of malignant struma-ovarii is difficult to predict because of scarcity of cases and the long intervals before recurrence or metastasis[7]. O'Connel *et al*[11] reported two cases of malignant struma-ovarii. One presented with malignant dissemination to retroperitoneal tissue and lymphglands. Other was of recurrence after ipsilateral Salpingo-Oophorectomy. Re-examination and evaluation of histopathological section of the original mass showed evidence of malignancy.

Salman *et al*[12] reviewed the literature and added one case of papillary thyroid cancer in struma-ovarii. The presentation is similar in the case reported here. In both histology of the mass established the diagnosis and years of follow up showed no evidence of recurrence.

Struma-ovarii is teratoma of the ovary where thyroid tissue predominates malignant variant of it is rare. The presentations are like that of similar ovarian tumours. Histology establishes the diagnosis of struma-ovarii and also the malignant change in it if any.

The prognosis also is good after conservative surgery Oophorectomy in malignancy in struma-ovarii when limited to the ovary and the capsule is intact.

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Conflict of Interest : Dr. Aruna Tantia, Dr. Sunipa Chatterjee, Dr. Shashi Khanna, Dr. Poonam Kapoor, Prof. M. S. Banerjee and Dr. Ushasi Mukherjee, declare that they have no conflict of interest and nothing to disclose.

IRB Details : ILS Hospital Ethics Committee Reg. No. – ECR/130/Inst/WB/2013/RR-19 (Validity – 20 April 2019 – 19 April 2024.) has declared on 05.03.2021 that this case report does not need any approval and is exempted for IRB approval.

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