When Thyroid Meets Ovary: A Case Report of Struma Ovarii

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ABSTRACT
Struma Ovarii is not a common tumor. It is a mature ovarian teratoma, completely or predominantly composed of thyroid tissue. It belongs to a specialized monodermal variety. Struma ovarii may have an association with hyperthyroidism; however, it is a rare association and just in about 8% of cases. Its diagnosis is mostly delayed because most of the patients do not present any symptoms. Some patients may develop symptoms of specific conditions like ovarian torsion, hyperthyroidism, and ascites. Ultrasound, MRI, CT scan, and scintigraphic evaluation are necessary to differentiate benign from malignant variety. The first line treatment option is surgical removal. There are lesser chances that the tumor will recur and the quality of life is improved significantly. In this case report, we have discussed a unique case of struma ovarii. It was duly diagnosed and appropriate management was done. The tumor was different from malignant ovarian tumor on the basis of ascites and tumor marker assessments. Thyroid function investigations were also normal. The tumor is incidentally found on imaging in most cases. The definitive diagnosis is established by a histopathological study. The first line of treatment is surgical removal and it usually leads to a successful prognosis. We will determine the challenges that are faced to reach the diagnosis and management of this rare tumor. Misdiagnosis is common. The treatment options are debatable because it is a rare tumor. In fertile and young age groups, fertility conserving surgeries like ovarian cystectomy or unilateral salpingo-oophorectomy are advised. For the purpose of treatment, in postmenopausal cases, total abdominal hysterectomy with bilateral salpingo-oophorectomy may sometimes be indicated. The quality of life will improve if investigations are done early and appropriate management is done.

Keywords: Ovarian mass, ovarian teratoma, pelvic mass, struma ovarii.

1. Introduction
Struma ovarii is mostly a teratoma. There is usually more than 50 percent of thyroid tissue present as a cellular component of this ovarian tumor. It is very rarely malignant. Struma ovarii commonly presents between the fifth and sixth decade of life and it makes for 3% of ovarian teratomas and 0.3% of all ovarian tumors [1].

Thyroid hormone secretion is uncommon; however, features associated with hyperthyroidism have been reported in 8% of cases [2]. The presentation is often with nonspecific symptoms and can be similar to ovarian malignancy. The diagnosis often comes as a surprise following surgery and when histopathological findings are reported. When the presentation is malignant, treatment is not clearly defined as it is a rare tumor [3]. The physician should be very careful when consulting the patient because the proper treatment and the prognosis are not very definitive due to their rarity. There are no symptoms in 40% of cases. These cases are incidentally identified during ultrasound. When symptoms become evident, they are usually tachycardia, abdominal pain, vaginal bleeding, palpable abdominal mass, and ascites [4].

2. Case Presentation
This case report describes a 26-year-old married nulliparous female patient who presented with complaints of on and off abdominal pain for 2 years and abdominal distension for 6 months. There was nothing significant
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Fig. 1. Solid cystic heterogeneous mass lesion of left adnexal origin.

Fig. 2. Doppler evaluation, which shows vascularity on Color Doppler scan.

Fig. 3. CT scan through lower abdomen reveal 14 × 13 cm heterogeneous solid cystic smooth lesion.

found in her past medical and family history. At the time of admission, her general physical examination was normal. But on abdominopelvic examination, a humped mass was felt in the pelvis. The size was about that of a tangerine. Her ultrasound scan revealed a heterogeneous rounded walled solid cystic mass arising from mid pelvis, which was thought to be adnexal as the left ovary was not separately visualized. Right ovary and uterus were unremarkable. Figs. 1 and 2 reveal the sonographic features of the lesion.

CT scan (Abdomen & pelvis) was done to further characterize the lesion. A 14 × 13 cm solid cystic heterogeneously hypo-dense lesion with central cystic (fluid density 29 HU) and peripheral solid component was noted with enhancement of solid component on contrast administration. No distant metastasis was appreciated in the liver, peritoneum, omentum, or visualized bones. CT scan features were contrary to the usual features of teratomas which show lipid contents or lipid-fluid or fluid-fluid levels. Normal right adnexal region with the normal uterus, moderate ascites, and small pleural effusion. Figs. 3 and 4 show CT scan features.

MRI pelvis was done to see the local extent of the lesion. It shows a fairly large centrally cystic peripherally enhancing lesion in the pelvis. The lesion was hypo-intense on T1WI, heterogeneously hyper-intense on T2WI with the solid component in the periphery, showing impeded diffusion in the solid component. On gadolinium, contrast-enhancement struma ovarii show a typical lacy pattern of contrast enhancement. MRI scan features of the lesion are shown in Figs. 5–7.

Keeping in view the young age, all germ cell tumor markers were advised and it was found that all Tumor markers were in the normal range, AFP was 1.46 (Normal 0–8 in non-pregnant females) B-hcG was < 1.2 (Normal 0–5), LDH was 130 (Normal 135–214) while Ca 125 was raised upto 2108 U/ml (Normal < 35) which gave the suspicion of epithelial carcinoma of ovary. To establish diagnosis, we advised ascitic fluid tap with cell block and apply IHC MARKER for definitive diagnosis. 120 ml yellow colour ascitic fluid was drained and a smear from the fluid revealed RBC, lymphocytes, rare macrophages, and mesothelial cells but no malignant cells were seen. The patient was young and desired fertility so we advised ultrasound-guided biopsy from the solid
Fig. 4. Hounsfield unit value of central cystic material is 29 HU. It can be up to 90 HU depending upon thyroglobulin contents in it. Note no fat attenuation is seen in the lesion. Ascites surround the lesion.

Fig. 5. Shows hypo-intense signals in the lesion—suggesting fluid signals and no fat signals.

Fig. 6. T2W axial imaging show heterogeneously hyper-intense lesion, with peripheral hypo-intense solid component.

component of the left adnexal mass. The core biopsy of the left ovarian mass shows atypical cellular proliferation with PAX8 positive, CAM5.2 positive, while synaptophysin SF1, CDX2, inhibin, SALL4 were negative. p53 wild type and CD10, WT1, and INSMI were Negative/Non contributory due to the scanty nature of the biopsy. The histopathology unit advised repeating a larger biopsy for a definite diagnosis. The second attempt of biopsy from the solid component of the left ovarian mass was done under CT scan guidance and it came out to be Struma Ovarii of monodermal origin. Meanwhile, patient CA 125 was raised to 2431 U/ml (Normal < 35) and her serum thyroglobulin was assessed considering the thyroid tissue in the teratoma, which also came out to be raised ~30,000 ng/ml (Normal < 55). Raised serum thyroglobulin also favored that ovarian mass contained thyroid tissue. The patient did not show hyperthyroidism in this whole time period before and after surgery. Fertility-sparing surgery was planned and the patient underwent removal of the left ovarian mass with left salpingoophorectomy, omentectomy, and peritoneal washing. Intraoperatively the uterus, right ovary, and fallopian tubes appeared normal and a smooth-walled rounded mass of the left ovary was noted with cystic elements. There were no palpable regional lymph nodes or adhesions with regional organs. Struma Ovarii of monodermal origin was reported on intraoperative frozen section biopsy. The uterus, right ovary, and right fallopian tube were looking normal. On microscopy, the growth pattern of the tumor was lobular and comprised of thyroid follicles resembling normal thyroid tissue. Cuboidal to columnar cells were noted, and a major component of the cell was with oxyphilic cytoplasm. Birefringent calcium oxalate crystals are seen in dense colloids of the follicles. The typical round to oval nuclei along with cytologic atypia and rare mitotic figures were revealed. There was a scanty intervening stroma and extensive edema. Post-op time was uneventful for the patient. Post-op serum thyroglobulin was 14 ng/ml (Normal < 55) and MRI pelvis showed no residual pelvic mass. Histopathological views are shown in Figs. 8 and 9.

3. Discussion

Struma Ovarii presents as different variants. The mature thyroid tissue appears in all of the peritoneal cavity in
the benign strumosis. On comparison, in carcinoid version which is malignant, the malignant tissue may be present within a Struma Ovarii [3]. Differential diagnosis of struma ovarii includes malignant epithelial ovarian neoplasm, endometriotic cyst/haemorrhagic cyst, and tubo-ovarian abscess [6]. The immature teratomas have small foci of fat, however, they are mainly solid. The mature cystic teratomas or dermoid cysts, on the other hand, are recognized to be mostly cystic. These present as a mass that contains fat. There is often an association between calcifications and an enhancing nodule-forming soft-tissue part [7]. The computed tomography (CT) and magnetic resonance (MR) imaging diagnosed it easily. These techniques are very sensitive to detecting fat within a tumor. The CT presents fat as a negative attenuation and MR scan presents as hyperintense on T1 and T2W imaging [8]. On MRI, there are three methods that differentiate the fatty contents of the mature cystic teratoma from endometriosis or other haemorrhagic cysts. Firstly, when fat is detected, there is a chemical shift artifact which is in the frequency encoding direction. It differentiates fat from haemorrhage [9]. Secondly, it can be done by gradient-echo imaging. It uses an echo time and the phase of fat and water are opposite. It demonstrates the fat-water interface and mixtures which consist of fat and water [10].

Thirdly, fat in teratoma is distinguished from haemorrhagic lesions as high signals of fat will be suppressed by certain sequences with frequency selective fat saturation. This technique is preferable due to its accuracy when compared with other techniques. The struma ovarii does not reveal lipid material on both CT or MRI when compared with other types of teratoma [11]. The non-functional Struma ovarii cannot be differentiated from other cystic masses by imaging. When there are no features of hyperthyroidism in struma ovarii, there may be another differential diagnosis, such as cystic teratoma which is mature and has no fatty component, endometriosis, cystadenocarcinoma, cystadenoma, tubo-ovarian abscess formation, and metastatic tumour. However, the images of such lesions may appear similar to struma ovarii [12].

The presentation of both mucinous cystadenoma and struma ovarii comes as locules containing hyperintense content on T1; it represents mucinous material in the mucinous cystadenoma and thyroglobulin in the struma ovarii. When the lesion presents with high-intensity locules on T1W imaging, a mucinous cyst should be given consideration. It is typical of mucinous contents and has septations. These features are also of struma ovarii, however, it is usually a more complex lesion compared to a mucinous cystadenoma. The mucinous cystadenoma does not reveal a lacy pattern of contrast enhancement. The very low signal on T1 and T2 due to gelatinous colloid in the struma may resemble the features of a mucinous tumor. It may be more similar to that of struma. The presentation of endometriotic cysts is as a very low signal intensity on T2-weighted imaging. The signal intensity on T1-weighted imaging is mostly high. It may be differentiated from struma ovarii by these features. In both benign and malignant cases, ascites is present. The clinical features of a tubo-ovarian abscess are pain, fever, and leucocytosis as compared to the more gradual presentation of struma. The differential diagnosis of ovarian cancer is more difficult. The features of malignant epithelial cysts whether serous or mucinous type, are very complex. It is nearly identical to the lacy pattern of struma ovarii. Moreover, when an ovarian cystadenocarcinoma has a haemorrhage, it resembles the colloid in struma. The differential from malignant epithelial cysts is also challenging if there is no omental disease.

The surgical resection of the ovary is the mandatory treatment. It is sufficient for benign struma ovarii. The total abdominal hysterectomy with bilateral salpingo-oophorectomy is sometimes indicated in post menopausal cases, where the aim is complete removal of the disease. The surgical outcomes should be followed and usually, the prognosis is excellent. The main clinical features in the present case were abdominal discomfort along with increased girth and mild shortness of breath. There was initial suspicion of abnormal malignant pathologies in the ovary in accordance with MRI and serum tumor markers. The definite diagnosis of struma ovarii was, however, made with further histological sampling and immunohistochemical study. The surgical management of the patients was successful and there was no evidence of recurrence.
4. Conclusions

There should be an appropriate treatment course. The possibility of misdiagnosed malignancy should be kept in mind. The treatment should be by an explorative laparotomy and the mass should be removed. There is also an option of fertility-sparing treatment; for which however patients should be selected carefully.

References


