Case Report

Proliferative struma ovarii: A rare case report

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Abstract

Dermoid cyst is the most common ovarian neoplasm, comprising up to 25% or more of all ovarian tumors, and contains various mature tissues derived from one or more of the embryonic germ layers, the ectoderm, mesoderm, and endoderm but occasionally transform to malignant. Struma ovarii is defined as teratoma containing predominantly thyroid tissue up to 50% of the cases. Stroma ovarii is a very unusual and rare type of teratoma and has been demonstrated in 5%–20% of the cases and comprises 3% of ovarian teratomas. For struma ovarii, patient age ranges between 6 and 74 years and most patients are in their reproductive years. Struma ovarii is predominantly unilateral and associated with mature cystic teratoma, ascites, pleural effusion, and rarely with a cystadenoma. Tumor is composed of mature thyroid tissue comprising varying sizes of follicles lined by columnar or flattened epithelium. Sometimes, the follicles are markedly crowded, but other criteria of malignancy are absent, and hence, these cases are reported as proliferative struma ovarii. Proliferative struma ovarii has a good prognosis. Some benign cases are misdiagnosed as carcinoma when serum CA-125 is highly raised. As there are no specific clinical, radiological, and serological markers available for this rare tumor, these are diagnosed on histopathology.

Keywords: Benign thyroid lesion, germ cell tumor, mature teratoma, proliferative struma ovarii

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INTRODUCTION

Struma ovarii is a rare histological finding, mostly occurring in reproductive age group. It comprises 3% of ovarian teratomas. Five percent to twenty percent of cases of mature cystic teratomas frequently contain thyroid tissue. Thyroid tissue represents the predominant component of tumor. Struma ovarii is mainly seen in the fifth decade and it can occur between the ages of 6–74 years. It presents with a palpable abdominal mass. Struma ovarii has a solid to cystic, soft and gelatinous, brown to green cut surface. An association with dermoid cyst or a mucinous tumor may be seen. Cases of struma ovarii with elevated CA-125 have rarely been reported. Struma ovarii rarely produces sufficient thyroid hormone to cause hyperthyroidism, or exceptionally become malignant, and thus may be managed as a thyroid cancer.[1]

CASE REPORT

A 45-year-old female patient presented with acute pelvic pain and abdominal fullness. Radiological investigation showed a diffuse, partially echogenic mass with multiple cystic and solid areas. The specimen submitted was that of

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total hysterectomy with bilateral salpingo-oophorectomy, specimen measuring 9x8x3cm, attached right ovary measuring 2x2.4x0.8cm, part of right fallopian tube measuring 3x0.8cm, left fallopian tube measuring 2.5x0.8cm part of right ovary measuring 10x4.5x4.5cm. Right ovary is large cystic tissue piece on cut shows large cystic areas filled with hair and tooth and few solid areas filled greenish brown gelatinous material. Bilateral fallopian tubes and left ovary were unremarkable. Microscopic findings showed a tumor having ectodermal component comprising squamous epithelium with appendages and mature glial tissue, mesodermal component composed of lobules of cartilage, and endodermal component composed of mucinous epithelium, many dilated thyroid follicles and many acinar/glandular structures were also identified [Figures 1 and 2]. Corpus luteal cyst was identified and the attached ovary also showed cartilage lobules and tiny inclusion cyst. Cervix showed a nabothian cyst. Endometrium was weakly proliferative endometrium and the sent part of Fallopian tube was unremarkable.

RESULTS

Based on the histology and immunohistochemistry, features were suggestive of mature teratoma (dermoid cyst) with proliferative struma ovarii. Immunohistochemistry for thyroid transcription factor-1 (TTF-1) is positive [Figure 3] and chromogranin, synaptophysin, CDX2, and pan-cytokeratin were negative.

DISCUSSION

Mature cystic teratoma (dermoid cyst) constitutes the majority of ovarian germ cell tumors and constitutes 20% of ovarian tumors. They are often discovered incidentally on physical or ultrasonographic examination. They may contain hair, teeth or bone, and fatty material. Thyroid tissue is rarely found on histological examination, but if the thyroid tissue predominates (>50%), then the term struma ovarii is applied. Struma ovarii, a very rare histological diagnosis, is found in just 3% of ovarian teratoma, 2% of all germ cell tumors, and 0.5% of all ovarian tumors. Malignant transformation is uncommon and occurs in only about 5% of struma ovarii. Most patients of struma ovarii are in reproductive age, but it can be diagnosed at any age, even in children. Most cases are clinically asymptomatic. It may be associated with ascites, with or without pleural effusion (Pseudo-Meigs syndrome). Macroscopically, the tumor is mostly solid or solid cystic, and sometimes cystic with solid areas or protrusions. The cut section may look greyish, with a fleshy glistening appearance due to thyroid component.[1,2] On microscopy, it is composed of mature thyroid tissue consisting of colloid containing follicles of various sizes. In the present case variably, sized

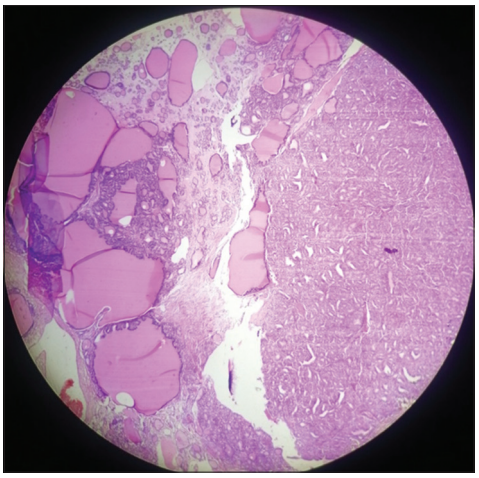

Figure 1: Varying sized thyroid follicles and crowded follicles filled with colloid and lined by benign follicular epithelium (H and E, ×10)

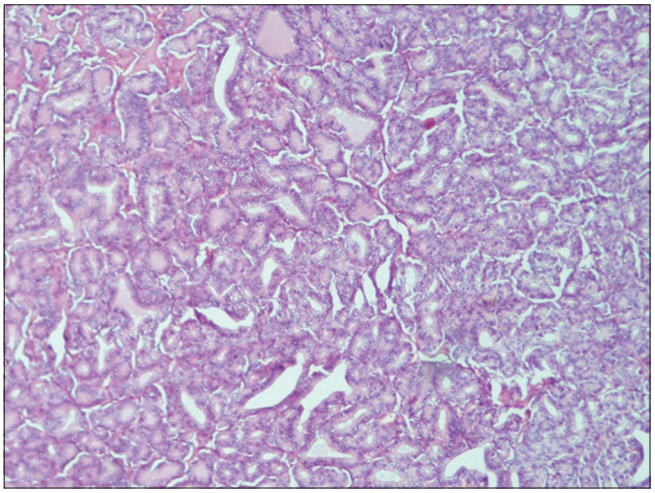

Figure 2: Follicles are markedly crowded, but without features of malignancy (H and E, ×20)

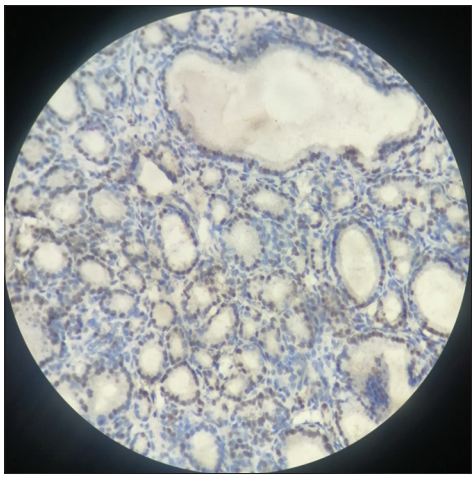

Figure 3: Thyroid follicles show Thyroid transcription factor-1 (TTF-1) nuclear positivity. IHC, X40

thyroid follicles filled with colloid were identified with foci showing marked crowding, but no proper capsule was identified excluding the possibility of follicular adenoma, also architectural features diagnostic of follicular thyroid carcinoma were not seen. Papillary architecture with papillary proliferation was seen, but nuclear features of papillary thyroid carcinoma (PTC) were absent in this case, so PTC was excluded and columnar cell variant of PTC was also excluded as CDX2 was negative. Histologically this case had morphology similar to carcinoid but was negative for synaptophysin and chromogranin excluding the possibility of carcinoid tumor. Most of the struma ovarii are benign and can be treated by salpingo-oophorectomy. Ultrasound is a primary modality for the identification and characterization of any ovarian mass. It is frequently associated with mature cystic teratoma or rarely with serous and mucinous adenoma. Most of the struma ovarii are solid or solid and cystic on macroscopic examination and predominantly or entirely cystic tumors and can lead to errors in diagnosis.[3,4] The epithelial lining of the cysts can have a largely nonspecific appearance composed of flattened cuboidal cells. Clues to the correct diagnosis include the identification of thyroid follicles in the cyst wall or in fibrous septa separating lobules of tumor and positivity for thyroglobulin and TTF-1.[4] Mature cystic teratoma shows focal high echogenic nodules with heterogeneous internal echoes. Typical feature of struma ovarii on sonography is the presence of well-defined solid tissue with a smooth margin that is vascularized on Doppler study ("struma pearl"). Proliferative Struma ovarii is benign, the retrospective studies of proliferative struma ovarii had good survival. Malignant changes in struma ovarii are the ones in which the tumor shows morphologically and histologically features of thyroid carcinoma. Benign struma ovarii and malignant forms without metastasis have a good prognosis. Ascites or pleural effusion if present disappears after surgery. Diagnosis must be made by pathological examination as a diagnosis by naked eyes intraoperative is not reliable. In pathological examination, especially during the frozen section pathological examination, the tumor including the capsule should be completely removed to avoid missed diagnosis. Struma ovarii containing thyroid type carcinoma must be distinguished from rare cases of papillary or follicular thyroid carcinoma metastasis to the ovary which is mainly characterized by cervical lymph node metastasis, and less commonly hematogenous metastasis.[4]

CONCLUSION

Struma ovarii can mimic ovarian malignancy clinically when it presents with a complex ovarian mass, with ascites and an elevated CA-125. Histopathologically diagnosed as proliferative struma ovarii. Proliferative struma ovarii has a good prognosis and survival without any significant long-term problems. There was no evidence of malignancy in all the slides reviewed. Efforts should be made to diagnose this condition preoperatively, to avoid extensive laparotomies, as these benign cases can be managed very effectively by the laparoscopic approach. Diagnoses are not straightforward, and pathologists should be cautious in diagnosing such lesions as benign because a malignant potential may coexist with innocuous histological features. In particular, even if a follicular proliferative lesion lacks definite malignant features, a diagnostic term recognizing low-grade malignant potential should be used.

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 initial s 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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