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Case Report

Struma ovarii: A rare entity

Moushami Singh[1], Raghu Ram Bhandary[1], Jitendra Pariyar[2], Swechha Maskey[1], Hari Prasad Dhakal[1]

[1]*Department of Pathology and Laboratory Medicine, Nepal Cancer Hospital and Research Center, Lalitpur, Nepal*
[2]*Gynecologic Oncology Unit, Nepal Cancer Hospital and Research Center, Lalitpur, Nepal*

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ABSTRACT

Struma ovarii, also known as goiter of the ovary is a rare disease.1 Considering the rarity of this tumor and constrained literature in Nepal's context, we hereby present a case of a 46-year-old female, asymptomatic, with normal blood parameters who presented with an abdominal mass. She was suspected of malignant ovarian mass on an ultrasonogram. Intraoperative frozen section examination revealed a benign tumor suggestive of dermal tumor versus struma ovarii. Postoperatively, histopathological examination was performed to assess the percentage of thyroid tissue and a diagnosis of benign struma ovarii was made which was confirmed by thyroglobulin positivity on immunohistochemistry. No features of immature teratoma and malignancy were identified.

Correspondence:
Dr. Moushami Singh
Department of Pathology and Laboratory Medicine
Nepal Cancer Hospital and Research Centre,
Harisiddhi, Lalitpur, Nepal
ORCID ID: 0000-0001-8647-3738
Email: moushamisingh123@gmail.com

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INTRODUCTION

Struma ovarii is a rare monodermal teratoma accounting for 2-4% of ovarian teratomas and 0.2- 1.3% of all ovarian tumors.[1,2] Thyroid tissue is nearly seen in 5-15% of dermoid tumors but to designate it as struma ovarii it should comprise more than fifty percent of mature thyroid tissue. The tumor is encountered in the fourth to sixth decade of life with reports being available from 6-74 years of age.1 Clinically, patients are usually asymptomatic while some present with an abdominal mass, pain, abdominal distension, and pelvic discomfort. Preoperative diagnosis with radiological findings using ultrasonogram (USG), computed tomography (CT) and Magnetic Resonance Imaging (MRI) is hardly possible.[1,2] Histopathological analysis provides an accurate diagnosis of benign struma ovarii.[3] Approach

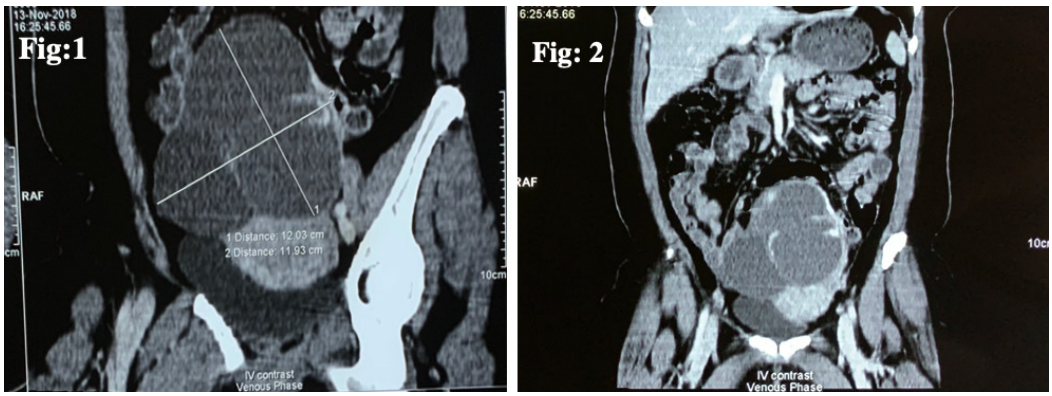

Figure 1 A and B: *CT scan of abdomen pelvis revealed presence of complex multiloculated cystic lesion in the left side of pelvis measuring 12.0 X 11.9 X 11.1 cm*

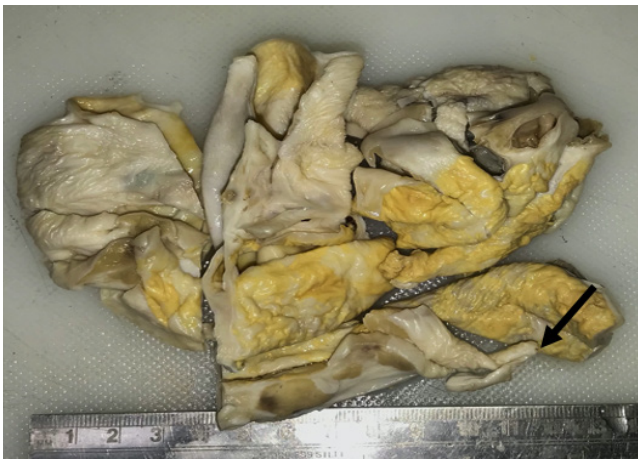

Figure 2: *Gross findings: Cut section revealing a solid and cystic component. The arrowhead shows the brownish gelatinous colloid*

to management is still controversial because of its rarity. Conservative surgery with fertility-sparing and follow-up is recommended for patients diagnosed with benign struma ovarii. Malignant struma ovarii may need adjuvant treatment and close follow-up since there is limited literature on its recurrence.

CASE REPORT

A case of a 43-year-old married, regularly menstruating, multigravida, Nepalese female who visited our hospital for routine medical examination. Her past medical and surgical histories were unremarkable. Her last childbirth was 17 years ago. Pap smear reports were negative for intraepithelial lesion and malignancy. On USG, a large left ovarian cyst measuring 12.0 X 7.5 cm was noted. Serum tumor markers, CA-125 (cancer antigen 125), and Carcinoembryonic antigen (CEA) was 13.4 ng/mL and 0.15 ng/mL respectively. CT scan of the abdomen pelvis revealed the presence of a complex multiloculated cystic lesion in the left side of the pelvis measuring 12.0 X 11.9 X 11.1 cm with internal enhancing nodular areas and extension in the midline and towards the left side. The lesion was extending and abutting the right ovary (fig. 1 A and B). The patient was counseled and scheduled for surgery: laparotomy and left salpingo-oophorectomy for intraoperative frozen section examination and further surgery based on the report. On intraoperative frozen section examination, left ovarian mass measuring 12x6 cm with intact smooth capsule was received. Cut section showed solid and predominantly cystic areas. The solid area was focally grey-brown. Representative sections from solid and cystic areas were submitted for examination during the frozen section (fig. 2). Microscopic examination showed bland benign-looking glands containing eosinophilic material within the lumen, which was in favor of benign thyroid follicles (fig. 3 A and B). A diagnosis of teratoma was thought of but considering the radiological findings which suggested malignant ovarian mass, well-differentiated adenocarcinoma was also ruled out by searching for areas of atypia, mitosis, and necrosis. Summing up the gross findings and microscopic findings, finally, a differential of teratoma versus struma ovarii was considered on frozen with a note for the examination of additional permanent paraffin blocks to assess the percentage of thyroid tissue. The operative procedure was limited to left salpingo-oophorectomy and right salpingectomy with omental biopsy.

Paraffin sections were prepared from multiple areas of grey-brown solid and cystic spots for further evaluation which revealed bland-looking thyroid follicles of variable size containing colloid (fig. 3A and B). No evident feature of immature teratoma and malignancy was identified in the sections studied. To further confirm the diagnosis immunohistochemistry with thyroglobulin was carried out. The thyroid follicular cells were highlighted by thyroglobulin immunohistochemistry. The histomorphological features combined with immunohistochemistry was consistent with struma ovarii.

The postoperative period was uneventful and the thyroid function test was within normal limits. A close follow-up was advised. There has been no recurrence or progression of the disease over one year period.

DISCUSSION

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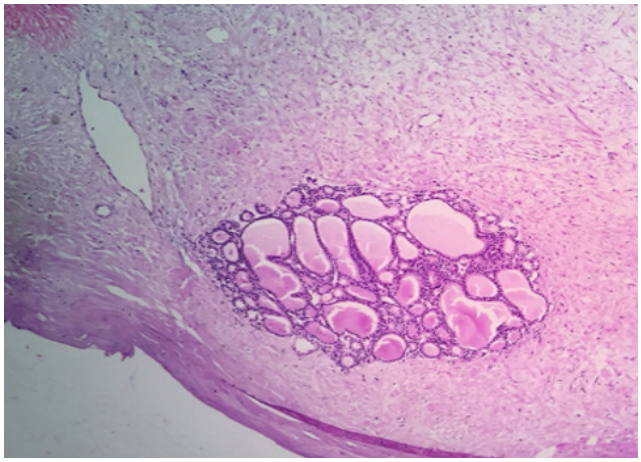

Figure 3A *Variable sized thyroid follicles containing colloid(HE stain; X40)*

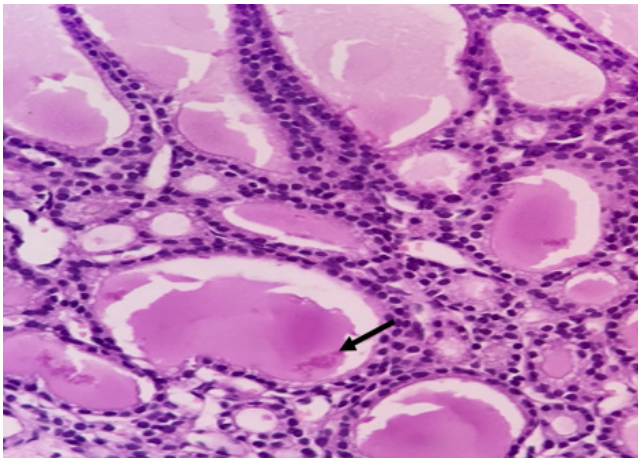

Figure 3B: *Variable sized thyroid follicles lined by benign follicular epithelial cells and containing colloid(HE stain; X400)*

Struma ovarii is a rare ovarian neoplasm, which was first reported by Boetllin in 1889.[5] Thyroid tissue is seen in 5-15% of ovarian teratomas but struma ovarii diagnosis is only designated when ectopic thyroid tissue comprises more than 50% of the overall ovarian tumor mass.[1,3,6] Incidence of this tumor is variable. In a study in Nepal carried out over 10 years from January 2006 to September 2015 by Ghartimagar et al, seven cases of benign struma ovarii were reported out of a total of 347 ovarian tumors whereas in a Japanese study, Higuchi et al, reported 3 cases out of 1000 solid ovarian tumors in 1960.[7,8] In our institute, this is the first reported case of benign struma ovarii out of an average of 600 cases yearly. Struma ovarii patients may present with palpable abdominal mass or laboratory findings of hyperthyroidism and increased radioiodine uptake (RAIU) in the thyroid gland. However, asymptomatic patients with laboratory studies within normal limits are diagnosed incidentally or during a routine abdominal and radiological examination as in our patient who was asymptomatic but suspected of malignant ovarian mass on the radiological study.[7] Radiological imaging studies can help identify multilocular cystic ovarian solid and cystic masses, but these do not allow differentiation of benign or malignant.

The ectopic thyroid tissue is inactive in most cases that result in normal thyroid function test. This is in concordance with our case. Patients with hormonally active thyroid tissue show hyperfunctioning struma ovarii based on the higher radioiodine uptake in the struma ovarii as compared to the thyroid gland.[8]

Ghartimagar et al reported 7 cases of struma ovarii in her study of which only 1 case was suggested as struma ovarii on imaging.[7] Pathologic examination is the gold standard in confirming the diagnosis of struma ovarii by analyzing the percentage of benign thyroid tissue and to differentiate benign from malignant struma ovarii.[9,10] As in our case, the patient was suspected of malignant ovarian mass on imaging studies which were later diagnosed and confirmed as benign struma ovarii on histopathological examination and immunohistochemistry. In the case presented above, the frozen section helped to identify the tissue as benign and to rule out malignant tumors of ovarian origin. The frozen section is a fairly simple test compared to paraffin sections. Paraffin sections are time-consuming but highly accurate. Although the diagnostic accuracy of the frozen section is high in ovarian masses, the false negative and false positive diagnosis should be taken into consideration to avoid misdiagnosis and understaging.[11] Most studies encourage the diagnosis of malignant struma ovarii based on the histopathological criteria of "ground glass" overlapping nuclei and nuclear grooves, or mitotic activity and vascular invasion.[4,6] However, based on histopathological findings of normal thyroidal tissue we were able to rule out malignant struma ovarii.

There is no standard consensus on the management of struma ovarii. Each case is managed differently relying on the patient's age, desire to have children, and whether it is benign or malignant. Treatment is limited to unilateral oophorectomy in patients desiring to have children.[4,9] Total thyroidectomy and adjuvant ablation are mandatory in the management of malignant cases.[12] Our patient had undergone laparotomy with left salpingo-oophorectomy, right salpingectomy, and omental biopsy. She is under regular follow-up and thyroid function tests are within normal limits.

CONCLUSIONS

Struma ovarii is a rare ovarian tumor. Patients may appear asymptomatic with normal thyroid function studies. Imaging studies such as CT scan and MRI may be suggestive of the morphology of the tumor but not diagnostic. Intraoperative frozen section is very useful in ruling out ovarian malignancy but for final conclusive diagnosis histopathological examination with immunohistochemistry with thyroglobulin of the tumor is required.

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