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Case Report

Pathology Section

Co-existence of Nodular Hyperplasia and Papillary Carcinoma Thyroid in a Case of Struma Ovarii: A Rare Occurrence

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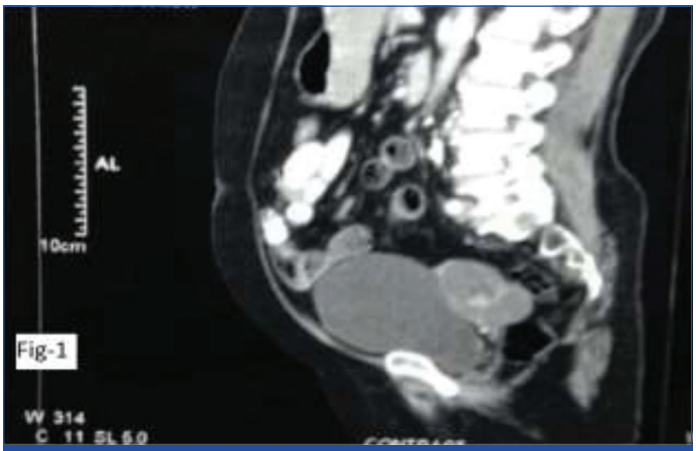
ABSTRACT

Struma ovarii is a monodermal ovarian teratoma. This is an ovarian germ cell tumour with more than 50% ovarian tissue. It is a rare tumour, accounting for only 3-5% of all ovarian teratomas. Malignant transformation occurs very rarely, only in about 0.3% cases. The authors report a case of struma ovarii with a malignant transformation to papillary carcinoma thyroid. Not much information is present in the literature about this rare entity. Due to the rarity of this condition, there is a lack of uniformity of histological criteria of malignancy and its management is not universally accepted by physicians. Here, the authors present a rare case of struma ovarii with malignant transformation to papillary carcinoma thyroid. A 62-year-old female presented with a left ovarian mass. On histopathological examination of the surgical specimen of hysterectomy it was found that the ovary had a neoplastic growth with morphological features of papillary carcinoma thyroid. The final diagnosis was struma ovarii with papillary thyroid carcinoma. Postoperative period was uneventful for the patient. However, follow-up was not done.

Keywords: Malignant, Monodermal, Ovary, Teratoma

CASE REPORT

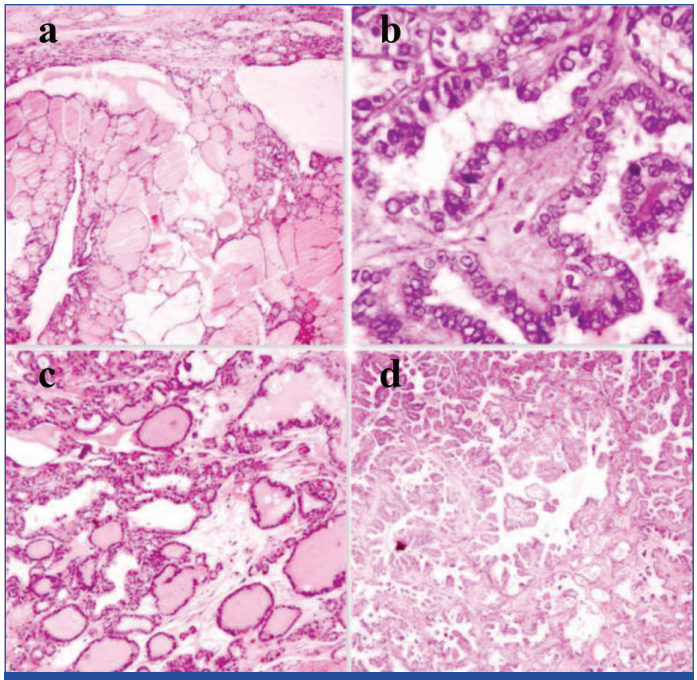
The authors present a case of a 62-year-old woman presenting with heaviness of lower abdomen for more than three months. Initially, there weren't any symptoms but gradually an ill-defined heaviness in the left lower abdomen developed with associated bloating and weight loss. On examination, no specific signs were present apart from a diffuse small swelling at the lower abdomen on palpation. Computed Tomography (CT) scan revealed a cystic solid left adnexal Space Occupying Lesion (SOL) with calcification measuring 6.0×5.7×5.4 cm, rest was within normal limit [Table/Fig-1]. The radiological diagnosis was left adenexal cystic SOL. Suspecting an ovarian tumour, her serum Cancer antigen 125 (Ca) level was done. It came out to be within normal range (2.6 IU/mL). Blood work-ups for thyroid were within normal limit. Other tumour markers like Carcinoembryonic Antigen (CEA) was 4.3, β-hCG was 3.55, Alpha Feto Protein (AFP) was 0.9 IU/mL. Fine Needle Aspiration Cytology (FNAC) was tried from the left adnexal SOL under image guidance but results were not satisfactory because adequate amount of material was not there in the smears. This was not sufficient for a conclusive opinion. After relevant work-up, a laparotomy followed by total abdominal hysterectomy with salpingo-oophorectomy with omental biopsy was done under spinal anaesthesia. The surgical specimen was sent to the Department of Pathology for histopathology report.



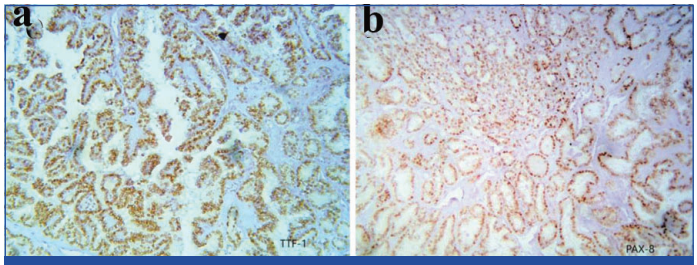
[Table/Fig-1]: CT scan of cystic solid SOL with calcification.

Composite specimen of uterus, cervix with B/L adnexa was received. A globular left ovarian mass was found measuring 5×4×2 cm with cystic consistency [Table/Fig-2]. On dissection, haemorrhagic areas with multiloculated cystic solid components were noted. Left sided fallopian tube was attached with this mass, other sided adnexa was unremarkable. Multiple relevant sections were taken from the specimen for tissue processing, paraffin embedding and Haematoxylin and Eosin (H&E) staining. Microscopic examination of sections from left adnexal SOL revealed ectopic thyroid tissue in the background of ovarian cytoarchitecture. This ectopic thyroid tissue was composed of more than 50% of the tumour parenchyma. This thyroid tissue showed features of focal nodular hyperplasia. Examination of more sections showed complex, randomly oriented branching papillae with fibrovascular core lined by cells with nuclear grooving, overlapping and optical clearing. Henceforth, the diagnosis of a monodermal struma ovarii with papillary thyroid carcinoma and focal nodular hyperplasia was framed [Table/Fig-3a-d]. Immunohistochemistry was done, which showed both nuclear positivity of Thyroid Transcription Factor-1 (TTF-1) and Paired Box Gene 8 (PAX8) that was further confirmatory of thyroid tissue in ovarian stroma [Table/Fig-4 a,b]. Postoperative period was uneventful. The patient was eventually discharged. In subsequent Outpatient Department (OPD) visits, the patient was in a good state.

[Table/Fig-2]: Gross appearance of the uterus, cervix and bilateral adnexal mass.



[Table/Fig-3a-d]: (a) Photomicrograph showing thyroid follicle in ovarian stroma (x100) H&E. (b) Photomicrograph showing papillary thyroid carcinoma in ovary (x400) H&E. (c) Photomicrograph showing nodular hyperplasia of thyroid in ovary (x100) H&E. (d) Photomicrograph showing papillary thyroid carcinoma in ovary (x100) H&E.



[Table/Fig-4a,b]: (a) Photomicrograph showing nuclear positivity of TTF-1 (x100). (b) Photomicrograph showing nuclear positivity of PAX8 (x100).

DISCUSSION

By definition, thyroid tissue in struma ovarii is more than 50% [1]. In 1888 the presence of ectopic thyroid tissue in an ovarian teratoma was first described by Bottlin, but struma ovarii as a separate disease entity was recognised 40 years later by the work of Ludwig Pick [2]. Malignant transformation is exceedingly rare. Only 5-10% of struma ovarii are histopathologically identical to differentiated thyroid carcinoma and recognised by the World Health Organisation (WHO) as Malignant Struma Ovarii (MSO) [3]. Not much is known about this rare entity. In a review by Marcy PY et al., it was stated that there are less than 200 cases of such in the literature [4]. Even after almost 100 years since its discovery, struma ovarii remains enigmatic, due to rarity of the condition. Malignant transformation of struma ovarii is even more rare. A number of cases reported as MSO in the past was stromal or insular carcinoid [5]. The authors present here a case of struma ovarii in a 62-year-old woman manifesting as a left ovarian cystic mass. As per the literature, the average age of struma ovarii is 43 years [6]. However, cases of struma ovarii in juvenile age of 10 years and in younger women (22 and 25 years) are also present. Most commonly occurring malignancy as per the literature is papillary carcinoma of the thyroid [7]. The histopathological diagnosis is mainly based on typical papillary pattern and nuclear features like empty ground glass appearance and nuclear overlapping [8]. In the present case, these microscopic features were clearly present. Nuclear features alone are not always sufficient for the diagnosis of malignancy; additional features like capsular invasion and peritoneal deposits are important as noted by Suzuki K et al., in their study on struma ovarii [9]. In the present case, peritoneal deposition as well as capsular rupture was not present.

However, there was strong nuclear positivity of TTF-1, which plays a cardinal role for the differentiation of thyroid follicular cells. It is also expressed in numerous tissues such as posterior pituitary and hypothalamus, in addition to thyroid [9].

As stated by other authors, PAX8, TTF-1 and Thyroglobulin (TG) immunohistochemical expression can be used to differentiate MSO from other ovarian neoplasms, but it is not clear whether these are of help in differentiating malignant from benign struma ovarii [10]. Sections from the tumour demonstrated strong nuclear staining for PAX8. PAX8 belongs to the paired box gene family, which is a important gene in organogenesis of several systems like thyroid, Mullerian system, and renal system. PAX8 positivity indicates its thyroid origin [11]. As stated earlier, relevant tumour markers were not contributory. CA125 is a marker for MSO but as per the literature, it is not effective due to its non specificity [10]. In this case CA125 was only 2.6 IU/mL corroborating with past reports. Whether histochemical markers like Human Bone Marrow Endothelial cell marker-1 (HBME-1), specific cytokeratins like Cytokeratin 19 (CK19), Rearranged During Transfection (RET) are of utility in diagnosis of MSO needs further investigation and research [12]. Due to the rarity of this entity, its immunohistochemcal profile has not been distinctively established. Marti JL et al., in their case series reported four cases of struma ovarii. Among these four cases, three cases had papillary thyroid carcinoma in the ovarian SOL. One case however had follicular thyroid carcinoma. They also reviewed 53 past reported cases of struma ovarii with well differentiated thyroid carcinoma. Among these 53 cases, papillary thyroid cancer was the most common histology (74%), followed by follicular variant of papillary (21%), and follicular thyroid cancer (5%). Therefore, these findings are corroborative with the present case. Metastasis was also not present in these cases, which was similar to the present case, where there was no evidence of metastatic involvement [13].

CONCLUSION(S)

Struma ovarii is a rare condition. As it is an elusive condition, not much information is present in the literature. Uniform histological criteria for malignancy are lacking. In the present case, after surgical excision the diagnosis was made as struma ovarii with a component of papillary thyroid carcinoma. Thus, diligent sampling of ovarian growth and careful microscopic examination is necessary for diagnosis. Further research is needed to identify the pathogenesis of this condition.

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