Struma Ovarii- Association with Pseudo-Meigs Syndrome and Raised S. CA-125: A Case Report

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ABSTRACT

Background: The monodermal mature teratoma known as Struma Ovarii is a rare and uncommon form of ovarian tumours. It is primarily composed of thyroid tissue and is associated with hyperthyroidism, increased levels of S. CA-125 (Cancer Antigen-125), and pseudo-Meigs syndrome. Because the symptoms of this tumour are nonspecific, other extremely frequent ovarian lesions may be misdiagnosed and ignored. The majority of the time, incidental imaging and symptoms are used to make the first diagnosis of the cancer; histopathology is used to confirm the diagnosis. A good prognosis and successful curative treatment can result from surgically removing the tumour.

The tumour's rarity makes misdiagnosis frequent. Nonetheless, a total abdominal hysterectomy combined with a bilateral salpingo-oophorectomy may be indicated in premenopausal patients in order to eradicate all symptoms. Ascites, hyperthyroidism, high S. CA-125 levels, and hydrothorax are all rare outcomes of struma ovarii. In this uncommon syndrome with negative cytology, individuals with ascites and pleural effusion should be examined for a differential diagnosis.

Keywords: Struma ovarii, Ovarian tumor, pseudo-Meigs syndrome, CA-125, Teratoma

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INTRODUCTION

Struma Ovarii is an uncommon type of ovarian tumour that primarily consists of thyroid tissue. It is formed from germ cell layers and manifests as a monodermal mature teratoma. Preoperative diagnosis is extremely challenging.[1] Even though the majority of tumours are benign, they can nonetheless mimic ovarian cancerous tumours. When a premenopausal woman has hydrothorax, ascites, and a multilocular mixed adnexal mass, elevated tumour marker levels should raise serious concerns about cancer.[2] Pathologists typically make the diagnosis after surgery. The disorder known as pseudo-Meig's syndrome, which is extremely rare, is linked to struma ovarii and increased levels of S. CA-125. This relationship makes it challenging to distinguish between malignancies.[3,4]

Many tumours produce thyroid hormone, and many tumour resections result in the development of hypothyroidism.[5] Although serous or mucinous cystadenomas are occasionally observed with this tumour, it is considered a developed teratoma.6 Less than 1% of ovarian tumours and 2 to 5% of all ovarian teratomas are included in this mass.[7] Due to the lack of specificity in the symptoms, it is frequent to misdiagnose them as other ovarian lesions.[8] The gold standard for treating cancer has been surgically excising the tumour and then administering adjuvant radioiodine therapy to prevent metastases and recurrence.[9]

The patient of this case report provided written informed consent and accompanying images for this journal's Editor-in-Chief to review and consider for publishing upon request. The reporting of this work complies with SCARE 2020.[10]

CASE PRESENTATION

A rare and unique case of a premenopausal woman who presented with benign struma ovarii associated pseudo-Meigs syndrome, elevated S. CA-125 ovarii, and a complex left pelvic mass that mimicked a malignant ovarian tumour based on assessments of tumour markers, recurrent pleural effusion, and ascites. The tumour was successfully diagnosed and treated. Thyroid function indices are normal, nonetheless.

A 41-years old woman, Rajina akter, housewife, (P3L3) referred to Combined Military Hospital (CMH), Dhaka in October 2023 with complaints of fever, weight loss, dyspnea and diffuses abdominal pain. On pulmonary auscultation, there was a decrease in the sound of the lungs on left side. On physical examination, abdomen was distended with fluid wave and dullness to percussion consistent of ascites without any evidence of per vaginal abnormalities. There was no evidence of lymphadenopathy or omental caking or other evidence of metastatic cancer. Baseline laboratory parameters including blood cell count, platelet count, as well as thyroid parameters (TSH, T4) were in the normal range. Her S.TSH, FT3 and FT4 were at 0.73 μIU/ml, 7.4 pmol/L and 12.1 pmol/L respectively. Laboratory tests revealed an elevated S.CA125 of >1000 U/mL of serum, whereas the remaining tumour markers were within normal limit. On Chest X-ray showed dense homogenous opacity with curvilinear upper margin extending along the lateral chest wall was noted on left side obscuring the left hemi-diaphragm & heart border that was evidence of marked left pleural effusions. CT scan of chest showed collapse, consolidation, inflammatory pulmonary lesions & gross pleural effusion on left side and tiny pleural based pulmonary nodules in right side. She underwent seven to eight times thoraco-centesis of effusion in left side due to re-accumulation rapidly of large amount of serous fluid that negative for malignancy and microorganisms.

Pleural fluid ADA was 5.3 U/L. The patient had no medical conditions including diabetes mellitus, hypertension, hypercholesterolemia, and thyroid disorders. The past medical history, drug history, and family history of the patient were negative. Firstly, assessed by abdomino-pelvic ultrasonography where large multilobulated mixed left adnexal cyst with thin septation, moderate ascites and collection in pelvis. Heterogeneous solid mass with the diameter of (70×58×67) mm & (51x53) mm with two thin-walled anechoic cystic regions in the left adnexa and severe vascularity in Colour Doppler examination (ORADS 5). Right ovary with the diameter of 22×25 mm was normal. On the basis of ultrasound, which concerned for malignancy, CT scan, MRI & further evaluation of tumour markers were performed. CT chest was also requested due to pulmonary symptoms. Assessing tumour markers indicated positivity for CA125 (>1000 U/mL) and ROMA Value (14.6) and negativity for HE4, S.CA19-9, βHCG, CEA and AFP. In MRI, uterus is normal. A large, measuring about CC (120×85×118) mm with peripheral calcification in one of the locule, enhancing mixed density abdomino-pelvic mass in left adnexa suggesting mitotic ovarian mass likely malignant ovarian teratoma. Mild ascites and omental infiltration and mild enhancement were also emerged. The findings were suggestive of malignant mass lesion in the left ovary. The right ovary was normal in size and other abdomino-pelvic organs were unmentionable (Fig-1 and Fig-2). CT Chest scanning revealed moderate unilateral (Lt) pleural effusion. To reduce the patient's pain and shortness of breath, pleural fluid was aspirated several times. Thoracotomy with drainage of pleural effusion was done to reduce breathlessness. The cytologic exam of the pleural fluid was negative for malignancy. The Pap smear test was also negative for intraepithelial lesion or malignancy. The patient underwent laparotomy. After exploration, revealing the removal of approximately 3 liters of ascites. A mass, adhering to the pelvic floor, was observed on the left side (Fig-3). The left ovary was sent to the laboratory for frozen section, leading to the conclusive diagnosis of struma ovarii in the left ovary (Fig-4). Omentum assessment showed inflamed congested adipose tissue without tumours. An endoscopic lung biopsy was conducted to rule out lung malignancy. Due to family completed, total abdominal hysterectomy with bilateral salpingo-oophorectomy were performed. The patient was discharged in good general condition after a 10-days. Subsequent follow-ups revealed normalized laboratory parameters, including thyroid function indices and S.CA-125 tumour marker. Thyroid ultrasonography showed no visible thyroid nodules.

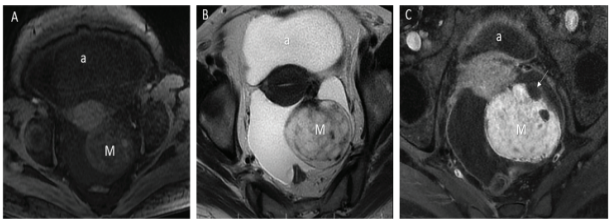

Fig-1(A) **Fig-1(B)** **Fig-1(C)**

Fig-1(A): Axial T1-weighted well-defined heterogeneous hypo-to isointense solid mass in left ovary. (B) In axial T2-weighted mass appeared hyper intense and contains multiple thick septa. (C) Post contrast T1-weighted fat-suppression manifested avid enhancement in solid component.

(a = ascites, M = mass, Arrow = follicule)

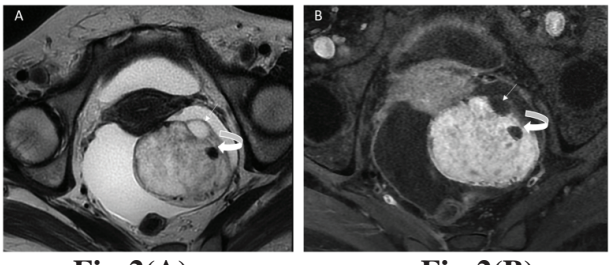

Fig-2(A) **Fig-2(B)**

Fig-2(A&B): T2-weighted and post contrast fat-sat T1-weighted showed a very low signal

intensity (curved arrow), due to the viscous colloid material that could be suggestive for the presence of struma ovarii tumor.

Thin arrow = follicule.

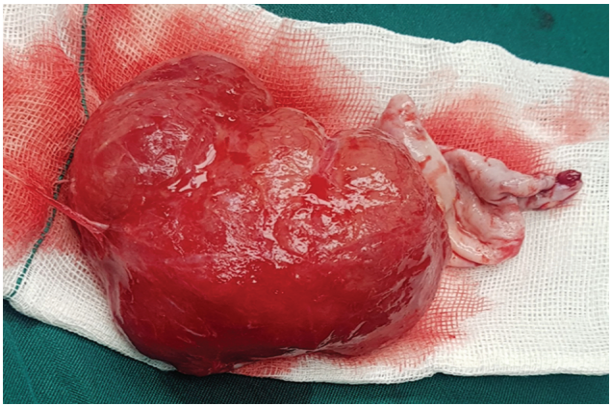

Fig-3: The feature of tumor resected surgically

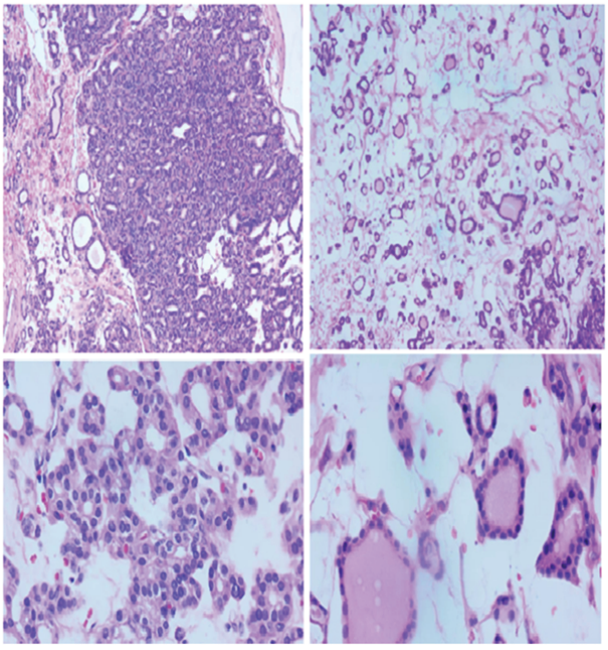

Fig-4: ×100 H & E staining feature and ×400 H& E staining feature

After six-month follow-up, the patient survived, and no mass was seen on imaging.

DISCUSSION

The peritoneal cavity may include mature thyroid tissue in benign strumosis, a rare variety of struma ovarii. Several kinds of struma ovarii have been reported. On the other hand, malignant tissue is present in the struma ovarii of the carcinoid variant.[11] Papillary thyroid carcinoma, which accounts for approximately one-third of cases and 5% of which show malignant metastases, is the most common malignant form.[12] Racial prejudice does not exist. It is rare for this tumour to manifest itself prior to puberty, usually occurring between the ages of 40 and 60.[13] Consistent clinical manifestations include ascites, palpable abdominal masses, irregular vaginal bleeding, and pseudo-Meigs syndrome (ascites with hydrothorax).[14] The tumour may be unintentionally found during pelvic imaging or surgery due to vague clinical symptoms. Additionally, 5-8% of individuals have thyroid dysfunction.[15] However, depending on its position and size, a physical examination may indicate a palpably enlarged abdominal mass. The more common gynaecological and abdominal diseases, such as ovarian cyst, endometrioma, thyrotoxicosis, ectopic pregnancy, and hydrosalpinx, must be carefully distinguished from stromia ovarii.[16] While S. CA-125 marker levels may rise in laboratory tests, haematological markers generally lie within the normal range and have limited specificity for this tumour.[17] Only when hyperthyroidism is symptomatic are thyroid function tests performed. When determining the extent of a disease and its connection with nearby organs, such as the colon, triple-contrast CT scanning is helpful.[18]

A multicystic tumour may show up on CT imaging with little to moderate cystic wall enhancement. When struma ovarii is pathologically present, it typically has a solid, greenish-brown appearance, although it can also have cystic features. Histological analysis reveals thyroid tissue as the main component, resembling a teratoma, and it is typically

present unilaterally.[19] The only essential and sufficient treatment for the benign variant of struma ovarii is surgical ovarian excision. Occasionally, for complete symptom alleviation in postmenopausal patients, a total abdominal hysterectomy with bilateral salpingo-oophorectomy may be advised.[20] An excellent prognosis is usually obtained with follow-up for surgical results. Abdominal pain, enlargement, and mild dyspnea were the main clinical symptoms in the particular instance that was given. There were no additional aberrant abdomino-pelvic findings. Based on MRI and serum tumour marker assessments, struma ovarii was initially considered to harbour aberrant malignant ovarian diseases. However, subsequent evaluation of histological samples along with IHC investigations definitively validated the diagnosis of struma ovarii. Following effective surgical therapy, the patient showed no symptoms of recurrence in later evaluations.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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