Case Report

DOI: 10.7860/JCDR/2022/60073.17283

Recurrent Right-side Pleural Effusion Mimicking Malignancy: Pseudo-Meig's Syndrome

Internal Medicine Section

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ABSTRACT

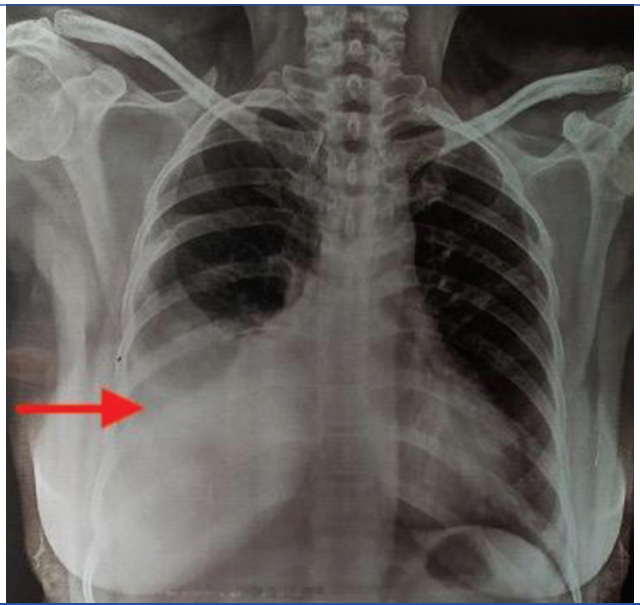
Pseudo-Meig's syndrome is an unusual presentation in which a benign ovarian tumour develops with ascites and pleural effusion. Diagnosis of exclusion is made only after ovarian fibroma has been ruled out and confirmed by disappearance of pleural effusion. The present case report is of a 55-year-old female, who presented with progressive shortness of breath, on and off right chest pain, and abdominal distension, over a period of 2-3 months. Clinical examination and a chest radiograph confirmed pleural effusion as the cause of progressive dyspnea. Presence of a pelvic mass and an elevated serum Cancer Antigen-125 (CA-125) increased the probability of malignancy. After complete tumour resection, a pathological report confirmed a benign ovarian tumour. The authors highlight the importance of suspicion, careful general examination, radiological evaluation, and histologic examination to confirm the diagnosis of pseudo-Meig's syndrome.

Keywords: Cancer antigen 125, Ovarian fibroma, Struma ovarii

CASE REPORT

A 55-year-old postmenopausal female presented with chief complaints of progressive shortness of breath, right sided chest pain on and off and abdominal distension past 2-3 months. She had a history of recurrent right-sided pleural effusion and underwent repeated thoracocentesis. The vitals were stable and she was maintaining SpO_2 98% on room air.

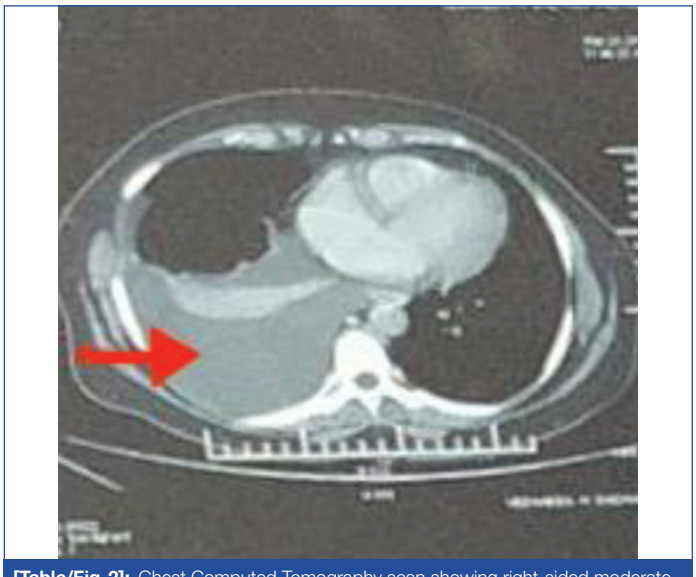
On auscultation, decreased intensity of breath sound was noted in right interscapular, infrascapular and infra-axillary areas. Chest radiograph Posteroanterior (PA) view showed right-sided homogenous opacity in mid and lower zone with right-sided Costophrenic (CP) angle blunted [Table/Fig-1].



[Table/Fig-1]: Chest radiograph, Posteroanterior (PA) view, suggestive of right side homogenous opacity in mid and lower zone with right-sided Costophrenic (CP) angle obliteration.

The patient was admitted, blood haemograms, Renal Function Test (RFT), Liver Function Test (LFT) were normal. Human Immunodeficiency Virus (HIV), Hepatitis B surface Antigen (HBsAg), Hepatitis C Virus (HCV) was non reactive and total serum protein was 6.1 gm/dL. Ultrasonography (USG) of thorax with abdomen and pelvis was suggestive of moderate right-sided pleural effusion, moderate ascites with hepatomegaly and heterogenous hypoechoic lesion of size 54×76 mm noted in right adnexa with cystic necrotic area within. A right adnexal neoplastic lesion with ovarian malignant neoplasm was likely.

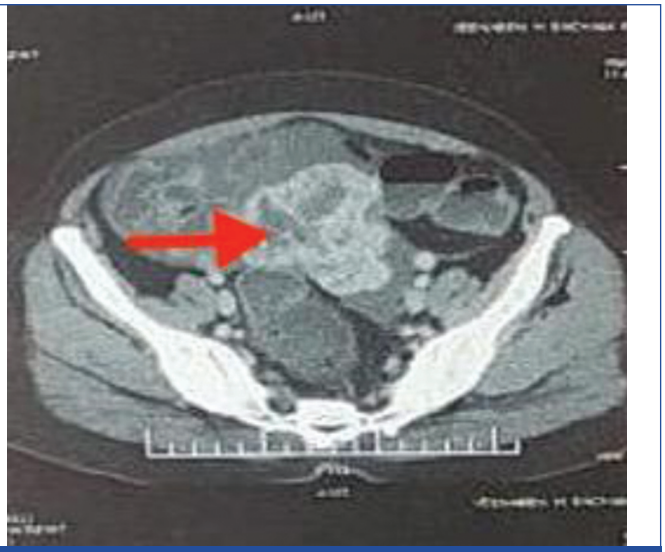
Contrast Enhanced Computed Tomography (CECT) abdomen and pelvis with thorax screening showed right-sided moderate pleural effusion [Table/Fig-2], moderate ascites with hepatomegaly and a well-defined heterogeneously enhancing soft tissue density lesion measuring 10×8 cm involving left ovary with cystic component within [Table/Fig-3]. Further blood investigations revealed, normal thyroid function test {Triiodothyronine (T3), Thyroxine (T4), Thyroid Stimulating Hormone (TSH)} but elevated Cancer Antigen-125 (CA-125) level (425 U/mL).



[Table/Fig-2]: Chest Computed Tomography scan showing right-sided moderate pleural effusion.

A therapeutic and diagnostic thoracocentesis and paracentesis were performed to alleviate symptoms- both pleural and ascitic fluids were sent for routine microscopy and cytology. The pleural fluid examination report suggested exudative pleural effusion according to Light's criteria (pleural fluid protein/serum protein) [1].

The ascitic fluid examination is detailed in [Table/Fig-4]. The Serum to Ascitic Albumin Gradient (SAAG ratio 1.1 g/L) points to the presence of exudative ascitic fluid.

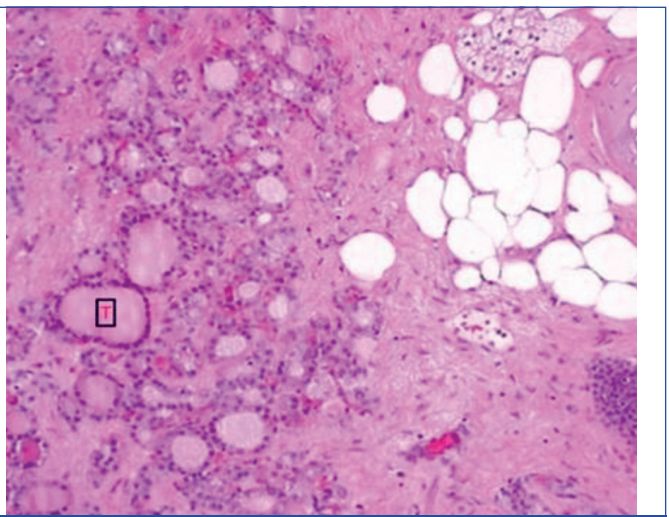


[Table/Fig-3]: Abdomen and Pelvis Computed Tomography scan showing well defined heterogeneously enhancing soft tissue density lesion measuring 10×8 cm involving left ovary with cystic component within.

Values	Pleural fluid examination	Ascitic fluid examination
Protein	5.0 gm/dL	6.7 gm/dL
Adenosine deaminase	20 U/L	15 U/L
Mesothelial cells	10%	25%
Lymphocytes	80%	65%
Total counts	600 cc/mm	300 cc/mm
Polymorphs	10%	10%
Impression	Exudative pleural effusion according to Light's criteria (pleural fluid protein/serum protein)	Exudative Ascitic fluid according to SAAG ratio (serum to ascitic albumin gradient <1.1 g/L)
Cytology	Not suggestive of any atypical cells.	Not suggestive of any atypical cells.

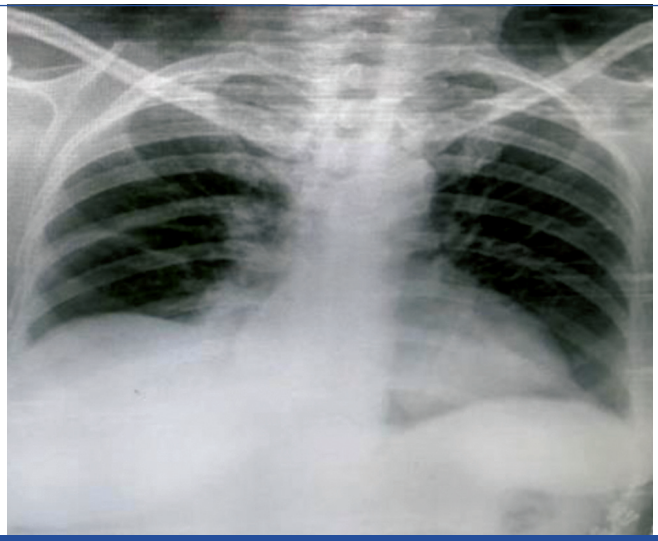
[Table/Fig-4]: Details of pleural fluid and ascitic fluid examination.

The patient was referred to a gynaecologist for further management. A total abdominal hysterectomy with bilateral salpingo-oopherectomy was performed. The left ovarian mass, sent for histopathological examination, was suggestive of mature cystic teratoma with dominant areas of struma ovarii showing thyroid follicles of varying size [Table/Fig-5]. The patient made an uneventful recovery and discharged on 10th postoperative day. After eight weeks on follow-up she was clinically and physically well with no evidence of diseases and normal CA-125 level (14.7 U/mL).



[Table/Fig-5]: Microscopic appearance of left ovary, showing thyroid follicles (T) of varying sizes. (H&E stain, 100X).

When she was examined a year later, she was still free of disease clinically and radiologically. Her follow-up chest radiograph posteroanterior view did not show any signs of pleural effusion, and her bilateral CP angles were clear [Table/Fig-6]. Her USG abdomen and pelvis revealed borderline hepatomegaly but no evidence of ascites.



[Table/Fig-6]: Chest radiograph, Posteroanterior (PA) view, showing normal lung field and with both side Costophrenic (CP) angle clear.

DISCUSSION

In 1937, Meigs and Cass first reported a series of cases in a patient with ascites, pleural effusion, and ovarian fibroma [2]. Meigs' syndrome is classically described as a triad of benign solid ovarian tumour with the gross appearance of a fibroma (either a fibroma, thecoma, or granulosa cell tumour) along with pleural effusion, ascites and resolution of effusion, ascites after tumour removal [3]. In contrast, pseudo-Meig's syndrome presents with ascites and pleural effusion, but with other pelvic masses [4] including mature teratoma, struma ovarii and ovarian, fallopian tube or uterine leiomyoma [5].

Meigs syndrome and pseudo-Meigs syndrome are very rare disorders. Meigs syndrome occurs in about 1% of all ovarian fibroids, uterine fibroids, and granulomas. Ovarian fibroid, ascites, and pleural effusion have been reported in 10-15% and 1% of cases, respectively [6].

Struma ovarii mimicking advanced ovarian cancer can be difficult to diagnose preoperatively [7]. Diagnosis of struma ovarii can simplest be made with the aid of using accomplishing histopathology [8]. This report focuses on a patient with struma ovarii who was initially thought to have a malignant tumour of the ovary before surgery based on clinical and radiological findings as well as elevated CA-125 levels. However, frozen sections and final histopathological reports showed a benign struma ovarii. In both syndromes, paracentesis and thoracentesis can be done to relieve symptoms, but treatment focuses on surgical removal of the tumour. If ascites and pleural effusion resolve and do not reaccumulate, a retrospective diagnosis can be made.

Struma ovarii mainly occurs during the 5th to 6th decade of life, with most cases being in postmenopausal women. The reported mean size of the tumour was 10 cm at its longest, mostly as a unilateral solid mass. They can sometimes be bilateral and contain cystic component. Malignant transformation of components is noted in 5-37% of struma, with metastases reported in less than 10% [9]. The index patient presented in 5th to 6th decade of life with an unilateral left sided ovarian lesion of size 10 cm in its largest dimension containing cystic component without metastasis.

The union of ascites, pleural effusion, and ovarian tumours in postmenopausal women with elevated CA-125 increases the likelihood of ovarian malignancy.

Even though there is a correlation between CA-125 and ovarian cancer, elevated CA-125 levels are commonly seen in some benign cases of Meigs syndrome. It is a non specific tumour marker and should not be used for the diagnosis of malignant ovarian tumours. There are other diseases associated with elevated CA-125 levels, including endometriosis, cirrhosis, pelvic inflammatory disease, and uterine fibroids. The exact reason for CA-125 elevation in Meigs and pseudo-Meigs syndrome remains unclear. One viable explanation is the subsequential irritation and inflammation of the pleural and peritoneal surfaces due to the presence of free fluid in these compartments [8]. In this report too the patient had an elevated CA-125 preoperatively which dropped down to normal after resection.

Several hypotheses has been proposed to explain the origin of ascites and pleural effusion in Meigs syndrome and pseudo-Meigs syndrome; although but it seems to be associated with lymphatic congestion but the exact cause remains unknown. Pressure from the tumour on lymphatic system can cause fluid to leak through the superficial lymphatic system [10,11].

A combination of intertumoral fluid leakage, mechanical irritation from the tumour, and inflammation of the peritoneum can result in ascites. For pleural effusions, mechanical transport of ascites via diaphragmatic orifices or lymphatics has been suggested [11-14]. Pleural effusions in Meigs syndrome and pseudo-Meigs syndrome have also been suggested to be on the right side. This is because the transdiaphragmatic lymphatics have a larger diameter on the right side. However, left-sided and bilateral pleural effusions have been reported [15]. Although ascites cytology can detect malignant cells in most of the cases, but has a high false-negative rate [16]. The index patient also presented with right-sided pleural effusion and ascites, but no cytologic atypia and no evidence of pleuroperitoneal connection in CECT Thorax.

Thyroid tissue is the major component of struma ovarii, and while various signs and symptoms of hyperthyroidism occur in 7% of patients, where on other side 25-33% have asymptomatic thyroid function test abnormalities [9]. After resection of benign struma ovarii Clinical hypothyroidism has additionally been reported [17]. In the present case, the preoperative thyroid function tests were normal, but the histopathology of the left ovary showed a mature cystic teratoma with dominant regions of struma ovaii showing thyroid cysts of varying size. However, there were no signs of hypothyroidism even after tumour resection has been noted.

The prognosis for this condition is excellent, with resection of the underlying tumour resolving both ascites and pleural effusions within a week. In the present case report, the patient's symptoms disappeared after surgery.

CONCLUSION(S)

Complete remission of ascites, hydrothorax and CA-125 occurred after surgery. Though, the combination of these cardinal features i.e, ovarian tumour, ascites and pleural effusion is highly suspicious for ovarian cancer in postmenopausal woman, rarely they can be of benign origin. Hence, Pseudo-Meig's syndrome should be considered in the differential diagnosis of patients presenting with suspicious clinical features but negative cytology.

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PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Sep 28, 2022
- Manual Googling: Oct 04, 2022
- iThenticate Software: Nov 28, 2022 (16%)

ETYMOLOGY: Author Origin

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Sep 05, 2022**
Date of Peer Review: **Oct 12, 2022**
Date of Acceptance: **Nov 29, 2022**
Date of Publishing: **Dec 01, 2022**