

Meigs' and Pseudo-Meigs' Syndrome in Patients Having Unexplained Pleural Effusion - A Case Series

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Abstract:

Introduction: Meigs' syndrome is characterized by the co-occurrence of ascites, pleural effusion and an ovarian tumor (fibroma or fibroma-like tumor). Pseudo-meigs' syndrome is associated with malignant or benign ovarian tumor (other than fibroma or fibroma-like) or uterine or fallopian tumors with similar triad. Meigs' syndrome is an uncommon occurrence, with only 1% of ovarian tumors being the cause.

Case Description: We are describing here in sequence one case of meigs' syndrome and two cases of pseudo-meigs' syndrome. First, second and third cases are in second, fifth and sixth decade of life respectively. Common clinical presentations were abdominal pain and shortness of breath. The clinical examination and various blood and radiological investigations were suggestive of raised CA 125, right sided pleural effusion, ovarian tumor with evidence of ascites in all three cases. Pleural and ascitic fluid tapping were done which were exudative in nature in all three cases. In first case 19 year old female, second case 41 year old female and third case 52 year old female, histopathology report of ovarian tumor were consistent with likely fibroma, bilateral serous cystadenocarcinoma and adenocarcinoma respectively.

Conclusion: Ascites, pleural effusion and elevated CA 125 in an elderly female raises the possibility of an ovarian tumor. Meigs' and pseudo-meigs' syndrome is typically diagnosed definitively after surgery, when the tumor is confirmed by histopathology and ascites and pleural effusion have resolved.

Keywords: Ascites, Cancer Antigen 125, Meigs' syndrome, Ovarian tumor, Pleural effusion, Pseudo-meigs' syndrome

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Introduction

Meigs' syndrome comprises of ascites, pleural effusion and benign ovarian tumor (fibroma, thecoma, Brenner tumor and rarely granulosa cell tumor)-which usually resolves after surgical removal of the tumor.^[1] Pseudo-Meigs' syndrome is the secondary accumulation of pleural effusion and ascites linked to ovarian tumors, either benign or malignant (not fibroma-like) or uterine or fallopian tumors with clinical signs like those of Meigs' syndrome.^[1] Meigs' syndrome is an uncommon occurrence, with about 1% of ovarian tumors causing it.^[2]

CASE 1:

A 19 year old, unmarried female came to emergency early morning with chief complains of shortness of breath on mild exertion associated with dry coughing since two days and abdominal pain since ten days, on and off, dull aching in nature associated with four to five episodes of vomiting containing food particles. No any significant past or family history related to presenting complains was there. On examination pulse rate was 108/min, respiratory rate was 28/min, SpO₂- 98% on room air, on chest auscultation right interscapular and infrascapular

breath sounds were reduced as compared to left. Chest X-ray PA view revealed blunting of right costo-phrenic angle suggesting of right sided pleural effusion (fig 1a). USG abdomen pelvis showed evidence of mild to moderate ascites with few internal septations. All routine blood investigations, renal function test, liver function test and electrocardiogram were insignificant. Contrast Enhanced CT Scan abdomen pelvis was done which showed large well defined lesion measuring

106×215×200 mm extending from right adnexa towards the epigastric region, right ovary was not seen separately from the lesion. The lesion was predominantly cystic with enhancing internal septations (fig 1b). Mild to moderate ascites noted. Few subcentimeter sized homogeneously enhancing Lymph nodes in pre-para aortic region also noted. Both pleural (fig 1c) as well as ascitic fluid tapping were done showing exudative nature of fluid in both.

Table 1: Analysis of pleural and ascitic fluid in case 1.

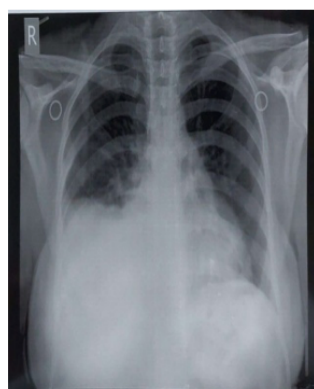
Characteristics	Pleural fluid	Ascitic fluid
Colour	Pale yellow	Pale yellow
Total Protein	4.9 g/dl	3.2 g/dl
Albumin	2.4 g/dl	1.8 g/dl
Glucose	105 mg/dl	90 mg/dl
WBC count	500/cumm	10/cumm
Polymorphs	10%	10%
Lymphocytes	90%	90%
Mesothelial cells	Seen in clusters and singly scattered/cumm	
ADA	12.61 U/L	10.2 U/L
Gram's stain	Negative	Negative
ZN stain	Negative	Negative
Aerobic culture	No bacterial organism isolated after 48 hrs of incubation at 37 degree celsius in ambient air.	

Table 2: Serum level of tumor marker tests.

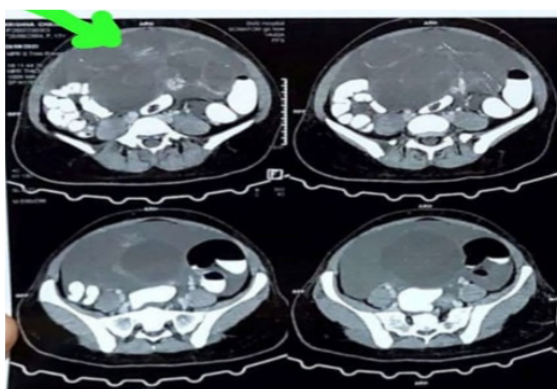
Test	Result	Reference range
S.CA-125	67 U/ml	<35U/ml
S. CEA	3.6 ng/ml	<7ng/ml
S. Alpha fetoprotein	3.4 ng/ml	<10ng/ml
S. Beta HCG	2.2 mIU/ml	<0.5mIU/ml

CA-125=Cancer Antigen 125, CEA = Carcinoembryonic Antigen, Beta HCG = Beta Human Chorionic Gonadotropin.

Figure 1:



1(a) Chest X-ray PA showing Right sided pleural effusion



1(b) Contrast Enhanced CT scan abdomen pelvis showing the large well-defined lesion.



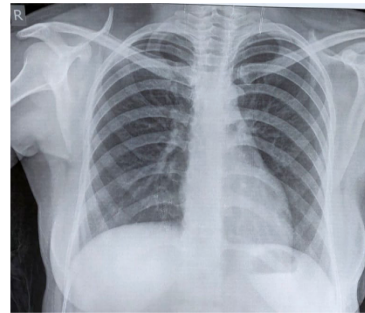
1(c) Pleural Fluid

Subsequently, the adnexal mass excision was done (fig 2a) and the ascitic fluid was drained intraoperatively. Postoperatively, the pleural effusion cleared within approximately one week (fig 2b). Histopathology of the mass revealed likely ovarian fibroma.

Figure 2:



2(a) Huge Adnexal Mass



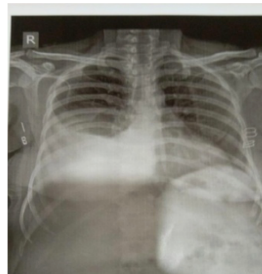
2(b) Post operative Chest Xray after one week

CASE 2:

A 41 year old female presented with abdominal pain since two months and shortness of breath on moderate exertion since one month. Patient was vitally stable and on respiratory examination right sided interscapular and intrascapular breath sounds were reduced as compared to left. All routine blood investigations and ECG were within normal limits. Chest X-ray revealed right sided pleural effusion (fig 3a). HRCT scan of thorax suggested of Gross pleural effusion on right side with underlying collapse of right lung. No any evidence of parenchymal infiltrate or mediastinal lymphadenopathy. Ultrasound abdomen pelvis suggested of mixed echogenic mass with internal

cystic component likely right tubo-ovarian mass. Moderate ascites was present. Contrast enhanced CT scan of abdomen with pelvis suggested of large 110×68×60 mm sized lobulated, heterogenously enhancing solid –cystic soft tissue mass lesion possibly arising from right adnexa. A serum CA-125 levels was performed which showed raised levels of 2284 U/ml. Both ascitic and pleural fluid tapping (fig 3b, 3c) were done which on investigations turned out to be exudative in nature. Total abdominal hysterectomy and bilateral salphingo-oophorectomy was done (fig 3d). Histopathological report of the biopsy revealed bilateral ovaries poorly differentiated serous cystadenocarcinoma.

Figure 3:



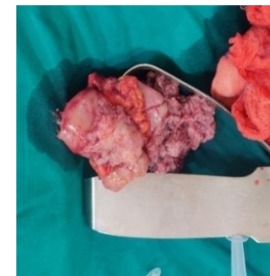
3(a) Chest X-ray showing Right sided pleural effusion



3(b) Ascitic fluid



3(c) Pleural fluid



3(d) Ovarian mass

Table 3: Analysis of pleural and ascitic fluid in case 2.

Characteristics	Pleural	Ascitic
Colour	Reddish	Reddish
Total Protein	5.1 g/dl	4.9 g/dl
Albumin	2.2 g/dl	2.4 g/dl
Glucose	113 mg/dl	108 mg/dl
WBC count	1000/cumm	200/cumm
Polymorphs	10%	18%
Lymphocytes	90%	82%
	Plenty of RBCs	Plenty of RBCs
ADA	9.43 U/L	13.2 U/L
Gram's stain	Negative	Negative
ZN stain	Negative	Negative
Aerobic culture	No bacterial organism isolated after 48 hrs of incubation at 37 degree celsius in ambient air.	

CASE 3:

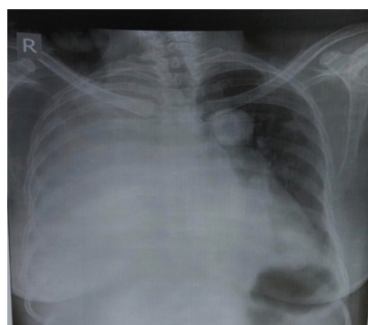
A 52 year old female presented to emergency department with chief complain of abdominal pain since 15 days, shortness of breath since three days, weight loss 4-5 kg in two months, dry cough since three days. No any known co-morbidities or significant past or family history was there. On examination, patient had tachypnea (Respiratory rate :28/min) and on respiratory examination right sided interscapular and infrascapular breath sounds were reduced as compared to left. Chest X-ray imaging revealed right sided hemithorax opacity with shifting of trachea to opposite side (fig 4a).

Ultrasound abdomen and pelvis revealed 40×30mm lobulate mixed echogenic lesion seen in right side of pelvis. Moderate vascularity is seen possibility of right ovarian mass. Moderate ascites present. Both pleural as well as ascitic fluid tapping (fig 4b,4c) was done which revealed exudative nature of fluid in both. Cytology in both fluids revealed presence of Adenocarcinoma cells. Serum CA-125 was more than 4000 U/ml. Positron Emission Tomography–CT scan showed irregular mass in bilateral ovaries, primary malignancy of bilateral adnexa (fig 4d). Metabolically active hypodense lesion in liver noted in segment five of right lobe.

Table 4 : Analysis of pleural and ascitic fluid in case 3.

Characteristics	Pleural	Ascitic
Colour	Reddish	Reddish
Total Protein	4.4 g/dl	3.5 g/dl
Albumin	2.3 g/dl	1.7 g/dl
Glucose	33 mg/dl	112 mg/dl
WBC count	300/cumm	6010/cumm
Polymorphs	70%	25%
Lymphocytes	30%	75%
	Plenty of RBCs	Plenty of RBCs Many mesothelial cells and atypical cells seen
ADA	15.6 U/L	7.2 U/L
Gram's stain	Negative	Negative
ZN stain	Negative	Negative
Aerobic culture	No bacterial organism isolated after 48 hrs of incubation at 37 degree celsius in ambient air.	

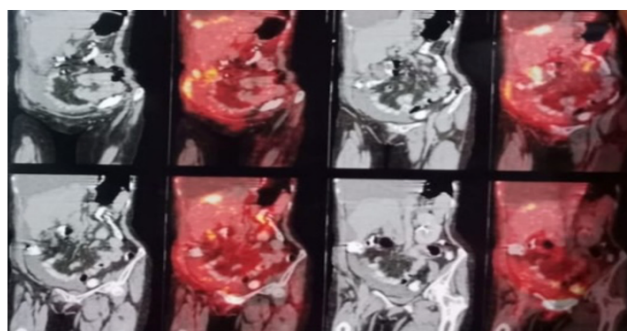
Figure 4:



4(a) Chest X-ray showing Right sided hemithorax opacity



4(b) Pleural Fluid



4(d) PET CT SCAN



4(c) Ascitic Fluid

Discussion

We describe here one case of Meigs' syndrome and two cases of pseudo-Meigs' syndrome. It is not possible to distinguish between Meigs' and pseudo-Meigs' syndrome from a clinical standpoint; so further pathological research is necessary.[2] Meigs' syndrome is extremely rare before to the third decade, but it gradually becomes more common after that, reaching its peak in the seventh decade.[3] In our case, we have three females belonging to second, fifth and sixth decade respectively. Ovarian fibroma patients experience ascites in 10% to 15% of cases, and hydrothorax in 1% of cases.[3] Right side pleural effusions account for about 70% of cases, left sided for 15%, and bilateral for 15%.[3] In our case, all three pleural effusions were right sided. The majority of Meigs' syndrome effusions (88.8%) are exudative in nature.[4] However, the

presence of a transudative effusion does not rule out this diagnosis.⁴ In our case series, all three pleural effusions were exudative. In first case 19 year old female, second case 41 year old female and third case 52 year old female, histopathology report of ovarian tumor suggestive of likely fibroma of ovary, bilateral serous cystadenocarcinoma of ovaries and adenocarcinoma of ovaries respectively. In women with malignant adnexal masses, the CA-125 cancer marker is frequently elevated.[5] But benign illnesses including endometriosis, uterine fibroids, pelvic inflammatory disease, menstruation, early pregnancy, pancreatitis or liver problems can also cause it to rise.[5] Meigs' and pseudo-Meigs' syndrome is typically diagnosed definitively after surgery, when the pleural effusion and ascites have resolved and the tumor has been confirmed histologically.[3]

Case	Symptoms of patient	Age (years)	CECT scan abdomen pelvis /PET CT scan (mass size)	Pleural Fluid and ascitic fluid Findings	Laterality of pleural effusion	CA-125	Histopathological examination Report
1	shortness of breath, abdominal pain, vomiting	19	106*215* 200 mm extending from right adnexa to epigastric region. Ascites +, pleural effusion +	Both are exudative	Right	Serum: 67 U/ml	Likely Fibroma
2	abdominal pain, shortness of breath	41	110*68*60 mm sized mass heterogeneously enhancing solid cystic soft tissue mass arising from right adnexa. Ascites+, pleural effusion +	Both are exudative	Right	Serum: 2284 U/ml.	Bilateral ovaries poorly differentiated serous cystadenocarcinoma
3	Abdominal pain, shortness of breath, weight loss, dry cough.	52	PET-CT scan revealed Irregular mass in bilateral ovaries, primary malignancy of bilateral adnexa, Metabolically active hypodense lesion in liver is noted in segment five of right lobe. Ascites+++ , pleural effusion +++ .	Both are exudative Cytology suggestive of malignant cells –Adenocarcinoma cells in both fluids.	Right	Serum Ca 125: >4000 Ascitic fluid CA 125: 11,089	Adenocarcinoma of ovaries

CECT abdomen pelvis: – Contrast Enhanced Computed Tomography scan

PET CT scan: –Positron Emission Tomography Computed Tomography scan

Conclusion

Pleural effusion, which is quite simple to identify clinically, frequently appears as the initial symptom of Meigs' and Pseudo-Meigs' syndrome in certain individuals. The workup should consist of a pelvic ultrasound, a CT scan of the chest, abdomen, and pelvis, pleural and ascitic fluid collection and analysis, and a serum marker of malignancy (CA-125). After the tumor is removed, pleural effusion

and ascites usually go away in a few weeks. If appropriately managed, both Pseudo-Meigs' syndrome and Meigs' syndrome have good prognosis.

Declaration of patient consent:

The authors certify that they have taken patients' consent. The patients had also given consent for case information to be reported in the journal for

educational and research purpose. Patients' name and identity will not be displayed.

References

1. Hou YY, Peng L, Zhou M. Meigs syndrome with pleural effusion as initial manifestation: A case report. *World J Clin Cases*. 2021 Jul 26;9(21):5972-5979.
2. Dalal N, Athwal PSS, Tharu B, Saravanan L, Mansour H. A Rare Case of Pseudo-Meigs' Syndrome With Ovarian Metastasis Presenting as Meigs' Syndrome. *Cureus*. 2020 Oct 18;12(10):e11022.
3. Saha S, Robertson M. Meigs' and Pseudo-Meigs' syndrome. *Australas J Ultrasound Med*. 2012 Feb;15(1):29-31.
4. Krenke R, Maskey-Warzechowska M, Korczynski P, Zielinska-Krawczyk M, Klimiuk J, Chazan R, Light RW. Pleural Effusion in Meigs' Syndrome-Transudate or Exudate?: Systematic Review of the Literature. *Medicine (Baltimore)*. 2015 Dec;94(49):e2114.
5. Iavarone I, Padovano M, Pasanisi F, Della Corte L, La Mantia E, Ronsini C. Meigs Syndrome and Elevated CA-125: Case Report and Literature Review of an Unusual Presentation Mimicking Ovarian Cancer. *Medicina (Kaunas)*. 2023 Sep 19;59(9):1684.