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# Pseudo Meigs' Syndrome

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## 1. Summary

Meigs syndrome is characterized by the presence of hydrothorax and ascites in the presence of solid, benign ovarian tumors that resolve after tumor removal. Pseudo-Meigs syndrome is defined as hydrothorax and ascites associated with pelvic tumors other than those identified in Meigs syndrome. Meigs syndrome is diagnosed on the basis of a triad of ovarian fibroid, pleural effusion, and ascites. A 48-year-old woman had abdominal distension and pain, and shortness of breath for one month. Hydrothorax, ascites, and a latero-uterine mass were discovered during clinical evaluation and paraclinical investigations. For symptomatic relief, a thoracocentesis was performed. An exploratory laparotomy was performed and the mass was diagnosed as an ovarian tumor of serous adenocarcinoma type, borderline after histopathological examination.

## 2. Introduction

Meigs syndrome is classically defined as the triad of ascites, pleural effusion, and benign ovarian fibroid. A key feature found in patients with Meigs syndrome is resolution of symptoms after tumor resection [1]. Meigs syndrome is a rare disease that can only be diagnosed after elimination of ovarian cancer. This remains a challenge because cancer antigen 125 (CA-125) is often elevated and workup findings usually reflect those of metastatic disease [2]. Meigs syndrome contrasts with pseudo-Meigs, which presents with ascites and pleural effusion in association with benign ovarian tumors (other than fibroids) and malignancies [3].

We present here a rare case of pseudo-Meigs syndrome mimicking Meigs syndrome at initial presentation.

## 3. Observation

H.B aged 48 years, mother of 3 living children, without any particular pathological history, was admitted to our department for chronic pelvic pain associated with an increase in abdominal circumference without other associated signs, all evolving in a context of alteration of the general state. The abdominal examination of the patient showed abdominal distension, a mobile dullness on percussion with a positive flow sign. Gynaecological examination revealed an enlarged uterus with no other associated signs. On respiratory examination, there was dullness on the left side with absent breath sounds. An abdominopelvic ultrasound was performed, which showed at the right latero uterine level the presence of a heterogeneous hypoechoic tissue formation of lobulated contours vascularized by Doppler measuring 6x5x6cm associated with a large peritoneal effusion (Figure 1).

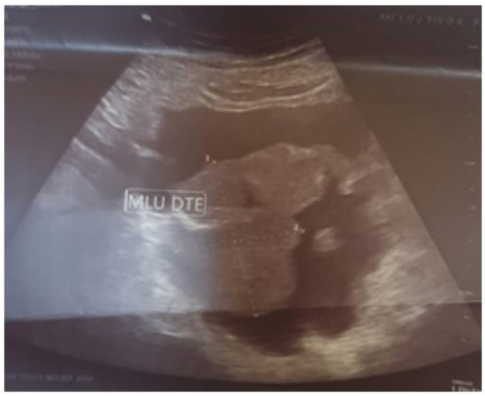


**Figure 1:** Ultrasonographic appearance of a right tissue MLU associated with a large effusion.

A pelvic MRI was ordered which revealed a right MLU of ovarian appearance measuring 5X4.5cm in T1 and T2 hyposignal enhanced after injection of PDC associated with a large amount of ascites of carcinomatous appearance with peritoneal nodules (Figure 2).

The chest X-ray revealed a left pleural effusion, with disturbance of the ventilation of the base, a blunt aspect of the right cul de sac. The patient underwent a thoracic CT scan which revealed a left

pleurisy with ascites of undetermined nature. A diagnostic thoracentesis was performed and 2 liters of cloudy yellow fluid were collected and sent for analysis. Fluid analysis revealed 900/mm3 of nucleated cells, 75% lymphocyte predominance, albumin 26 g/l, and a positive rivalta test confirming the exudative origin of the fluid. The cytology result was negative for malignant cells. CA 125 was highly elevated at 3550 U/ML.

The patient underwent an exploratory laparotomy with the presence of a large ascites with a 6 cm right MLU and thick peritoneal granulations, an epiploic cake and hepatic nodules. Biopsies of the MLU, peritoneal and epiploic with cytological sampling of the ascites fluid were performed.

The biopsy of the latero-uterine mass revealed a morphological aspect of a borderline serous adenocarcinoma with high grade infiltrating foci and presence of vascular emboli. The peritoneal granulation biopsy showed a peritoneal location of a moderately differentiated and invasive adenocarcinoma and the epiploic biopsy revealed an epiploic location of a moderately differentiated and invasive adenocarcinoma. The study of the ascites fluid showed the presence of carcinoma cells. The postoperative course was simple. The patient was referred to the oncology-radiotherapy department for further therapeutic management.

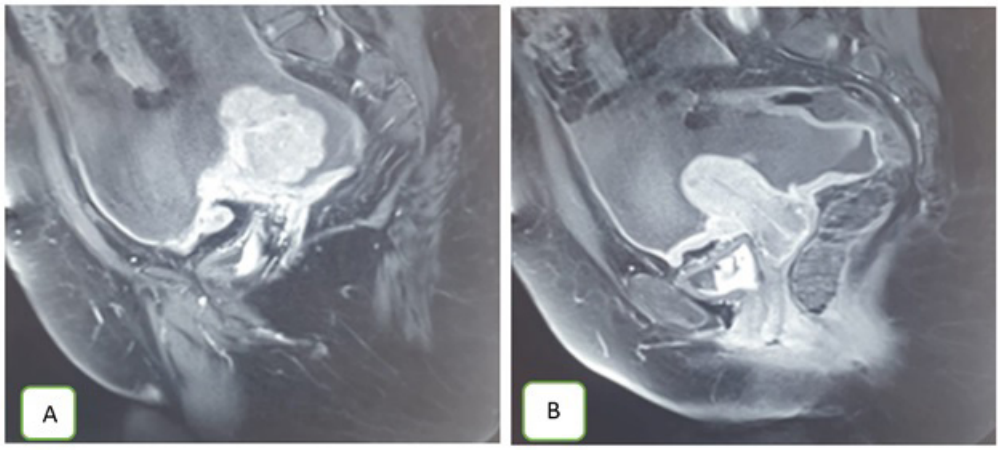


**Figure 2:** A And b Right MLU of ovarian appearance measuring 5X4.5cm enhanced after PDC injection associated with abundant ascites

## 4. Discussion

The association of an adnexal mass with pleural effusion and ascites in our case made us suspect Meigs syndrome. This syndrome is a rare disease characterized by pleural effusion and ascites in patients with ovarian fibroid or fibroid-like tumors [4]. It was first described in 1887 by Demons and then in 1937 by Meigs, who reached the same conclusions about the association of pleural effusion, ascites and benign ovarian fibroid. Therefore, this syndrome is also known as Demons-Meigs syndrome [5]. On the other hand, benign ovarian tumors (other than fibroids) and malignant ovarian tumors, such as mature teratomas and struma ovarii, can also be associated with pleural effusions and ascites, but they are classified as pseudo-Meigs syndrome [3].

Krenke et al. reviewed the characteristics of pleural effusion in reported cases of Meigs syndrome. They found that the average amount of pleural fluid collected was about three liters and that the majority of pleural effusions usually occurred on the right side. In our case, the pleural effusion was on the left side. The majority of pleural effusions in patients with Meigs syndrome are exudative [6]. In our case, pleural fluid analysis also revealed an exudative effusion without malignant cells, consistent with the majority of cases of Meigs syndrome. Review of reported cases of pseudo-Meigs syndrome shows that the pleural fluid in these patients contains reactive mesothelial cells without neoplastic cells [7].

Pleural effusion in Meigs syndrome is thought to be secondary to the passage of ascites fluid to the pleural space through the diaphragm or diaphragmatic lymph nodes [5]. It has been suggested that because the transdiaphragmatic lymph ducts are larger in diameter on the right side, the pleural effusion in Meigs syndrome "and pseudo-Meigs syndrome" is also on the right side. However, left-sided pleural effusions and bilateral pleural effusions have been reported [8]. It may present as a right pleural effusion with ascites, as seen in our patient, although rarely a pericardial effusion has been reported in the past and called Meigs-type syndrome [9]. Several hypotheses have been proposed for the underlying pathophysiology of ascites in these patients. Fluid leakage from edematous fibroids, tumor pressure on the pelvic and abdominal lymphatics, and causing lymphatic blockage are among these theories [10]. Vascular endothelial growth factor (VEGF), which increases capillary permeability, is also thought to be associated with pleural and peritoneal fluid formation. Ishiko et al. reported a significant difference in VEGF levels in pleural and peritoneal fluid before and after tumor resection in patients with Meigs syndrome [11].

CA-125 antigen is a tumor marker associated with ovarian carcinoma. Nevertheless, high levels of CA-125 have also been reported in the literature for Meigs syndrome, although levels above 1000 U/mL have been rarely reported [2,12].Lin et al. used immunohistochemical techniques to localize CA-125 expression, and

found that it is expressed by the mesothelium rather than the fibroma [13]. Case reviews have shown that higher levels of CA-125 are associated with higher ascites volume, but tumor size was not linearly correlated with CA-125 levels [14].

Diagnosis involves ruling out other conditions on the basis of pleural fluid/ascites analysis and imaging. A pseudo-Meigs syndrome has been reported in patients with systemic lupus erythematosus, which should be excluded by appropriate serological testing [10].

Removal of the tumor will ultimately result in resolution of ascites, pleural effusion, and normalization of CA-125 in Meigs and pseudo-Meigs syndrome [1,15]. However, surgery may not be a feasible option for all patients. Patients with malignant pleural effusion and ascites or those with significant comorbidities will not always choose to pursue resection of the adnexal mass. This is where symptom control and palliative medicine are sought. Repeated large-volume paracentesis and/or thoracentesis have been palliative choices for these patients. Another option is placement of an abdominal indwelling peritoneal catheter, which is associated with the same complication rates but reduces separate patient encounters and increases patient satisfaction [16]. Repeat thoracentesis, indwelling pleural catheter (IPC), and pleurodesis are the available therapeutic modalities for the management of symptomatic pleural effusion. Numerous studies have reported that IPC placement not only provides good symptom control, but can also lead to spontaneous pleurodesis [10,16] . Therefore, it is important that providers counsel patients with Meigs syndrome who are not ideal candidates for surgery on all available palliative options for symptom relief.

## 5. Conclusion

Meigs syndrome and pseudo-Meigs syndrome cannot be differentiated clinically and require further pathological studies. Although used to monitor therapeutic progress, serum CA125 levels are a poor biochemical marker used in the diagnosis and differentiation of malignant versus benign ovarian mass. This is a unique case of pseudo-Meigs syndrome mimicking Meigs syndrome at presentation, but upon further investigation, it turned out to be an ovarian adenocarcinoma. Cancerous processes should be suspected even in young women of reproductive age with a non-gynecological clinical presentation, early examination, and an adequate comprehensive approach for a better quality of life in this group of patients.

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