Meigs syndrome and elevated CA-125 levels, clinical mimicry with ovarian cancer. A case report

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Case Report

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Background: Meigs syndrome is a rare gynecological condition characterized by the triad of a benign ovarian tumor (commonly fibroma), ascites, and pleural effusion, all of which resolve after surgical removal of the tumor. First described by Meigs in 1937, this syndrome occurs in only 1–2% of ovarian fibroma cases. Although CA-125 is typically associated with epithelial ovarian cancer, mild to moderate elevation can be observed in Meigs syndrome due to factors such as peritoneal irritation and mesothelial cell activity.

This case report presents a 47-year-old woman with progressive abdominal distension and dyspnea. Imaging revealed a large right ovarian mass, ascites, and a right pleural effusion. CA-125 levels were significantly elevated at 565 U/mL. Following a radical hysterectomy with right oophorectomy, histopathology confirmed a benign ovarian fibroma. Postoperatively, the patient's ascites and pleural effusion resolved completely.

Meigs syndrome should be considered in the differential diagnosis of patients presenting with ascites, pleural effusion, and an ovarian mass, even with elevated tumor markers like CA-125 since these findings do not always indicate malignancy. Understanding this rare syndrome is essential, as surgical removal of the benign tumor results in full resolution of symptoms and favorable outcomes, avoiding unnecessary aggressive oncologic treatment.

Keywords: Meigs syndrome, CA-125

Meigs syndrome is an extremely rare gynaecological disease, presenting as fibroma or a fibroma-like tumor (thecoma, granulosa cell tumor, or Brenner tumor) accompanied by ascites and hydrothorax that rapidly resolve after removal of the tumor.[1] This syndrome was first reported by Joe Vincent Meigs in 1937 in a series of 7 cases where patients presented with an ovarian fibroma and associated ascites and hydrothorax.[2] Ovarian fibromas constitute the majority of the benign tumors seen in Meigs syndrome.[3] Fibromas represent approximately 4% of ovarian tumors and Meigs syndrome occurs in only 1% to 2% of these cases, thus is a rare condition.[4] CA-125 (cancer antigen-125), is a human glycoprotein employed for the diagnosis and follow-up of different cancer histotypes: first, epithelial ovarian cancer.[5] Commonly, it is slightly raised in Meigs Syndrome, although in the scientific literature, a value above 1000 IU/mL is rare.[6] The present work is a case report of Meigs Syndrome with elevated CA-125 levels.

Case report

A 47-year-old woman with no chronic illnesses, G2 P2, and no history of smoking or alcoholism, began experiencing an increase in abdominal circumference three years ago. Two months ago, she presented with progressive dyspnea, which occurred with moderate to mild exertion. A thoracoabdominal computed tomography scan showed right pleural effusion with right lung collapse **(Figure 1),** a complex right ovarian mass measuring 20 x 17 cm with free fluid**,** and an image suggestive of a Sister Mary Joseph´s node **(Figure 2)**. During the hospitalization, 3 thoracocentesis evacuators were performed, extracting a total of 4 liters. Tumor marker levels were: CEA 1.6 ng/mL, reference levels <5.0 ng/mL, hCG <1.2 mIU/mL, Ca 19-9 5.2 U/mL, reference levels <37 U/mL, AFP 2.6 ng/mL, reference levels 0.89-8.78 ng/mL, CA-125 with elevated levels of 565, reference levels of <35 U/mL.

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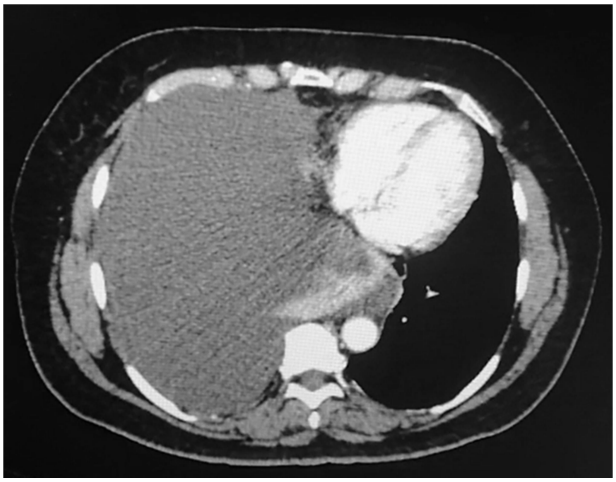

Figure 1. Chest CT scan showing right pleural effusion.

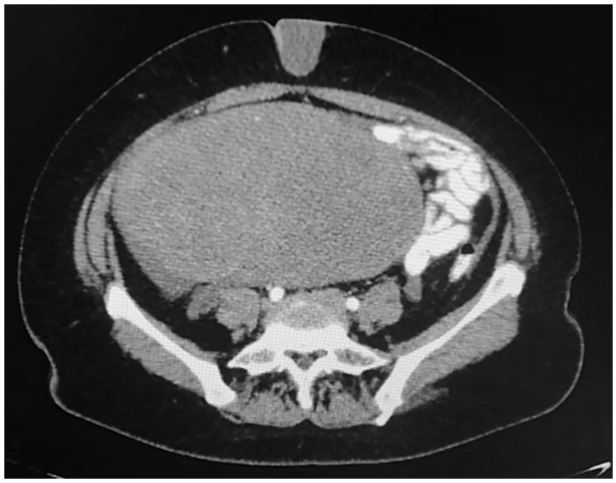

Figure 2. Abdominal CT scan showing a right ovarian mass.

The Oncosurgery service performed a radical hysterectomy plus right oophorectomy, revealing a right ovarian tumor **(Figure 3) (Figure 4)**. The 1,027 g ovarian sample is sent to histopathology characterized by round, spindle-shaped cells within collagenous stroma. After the surgical procedure, the patient's pleural effusion and ascites resolved and is currently being monitored by the Oncosurgery service.

Discussion

Meigs syndrome is diagnosed with the triad of ascites, pleural effusion, and an ovarian tumor, which is usually benign, occurring together.[7] Demons described a syndrome based on nine patients with benign ovarian cysts who were cured of their ascites and pleural effusion by removal of the cyst.[8] However, this syndrome was not named until 1937 when Joe Vincent Meigs along with Cass published a series of seven patients presenting with a triad of findings: Ascites, hydrothorax, and fibroma of the ovary characterized by the resolution of symptoms with ablation of the tumor.[7] Pericardial effusion is not included in the definition of Meigs syndrome; however, there have been case reports of patients with unexplained persistent pericardial effusion that resolved after the removal of a benign ovarian tumor.[9]

Meigs syndrome is diagnosed in association with approximately 1% of ovarian tumors, most of which are found to be benign ovarian fibroma on final histologic analysis.[4] About 10% to 15% of women with ovarian fibromas have ascites, and 1% have hydrothorax.[10] Approximately 70% of pleural effusions are right-sided, 15% left-sided, and 15% are bilateral.[11] The etiology of ascites and pleural effusion in Meigs syndrome is subject to debate and largely remains to be elucidated. There are several hypotheses regarding the mechanism underlying the generation of peritoneal fluid. It probably occurs by means of a transudative mechanism through the surface of the tumor that exceeds the resorptive capacity of the peritoneum.[12] Other potential explanations include hormone stimulation, obstruction of lymphatic flow by the tumor, and release of inflammatory cytokines and growth factors by tumor cells. The direct cause of pleural fluid formation is thought to involve translocation of ascites to the thoracic cavity via diaphragmatic pores.[13]

The most common histological types in the scientific literature are cellular fibroma, fibro-thecoma, fibroma, thecoma, and granulosa cell tumors.[6] Although various cases of Meigs Syndrome with elevated CA-125 serum levels have been described over the years, only 1% of ovarian fibromas or fibrosarcomas have presented with ascites and pleural effusion. For that reason, Meigs syndrome is considered rare in that specific context.[8,14]

CA-125, commonly used as a tumor marker, is a strong independent prognostic factor for epithelial ovarian cancer.[15] Despite the sensitivity of CA-125 in the detection of ovarian cancer, its specificity is known to be suboptimal. Many other conditions (neoplastic or non-neoplastic) can also cause an elevation of CA-125 levels, including endometriosis, cirrhosis, uterine fibroids, pregnancy, ovarian cysts, and pelvic inflammatory disease.[16]

In a determination of CA125 levels of 988 women with benign gynecological disorders, Niloff et al., observed the CA125 level to be more than 65 IU/ml in 1% on a single determination and only 0.5% on two sequential determinations.[17] Other reports of CA125 assessment in patients with benign stromal tumors without ascites describe levels less than 35 IU/ml.[18] However, in patients with ascites with or without hydrothorax, CA125 levels are much higher. Notably, the exact mechanisms that lead to CA-125 elevation in patients with Meigs syndrome remain unclear. Some studies have shown that mesothelial cells from ascites in patients with Meigs syndrome are able to synthesize CA-125.[19]

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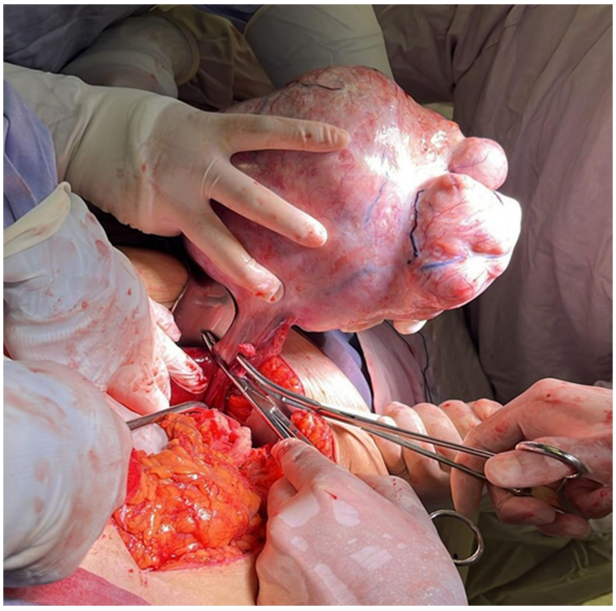

Figure 3. Intraoperative view showing the right ovarian tumor.

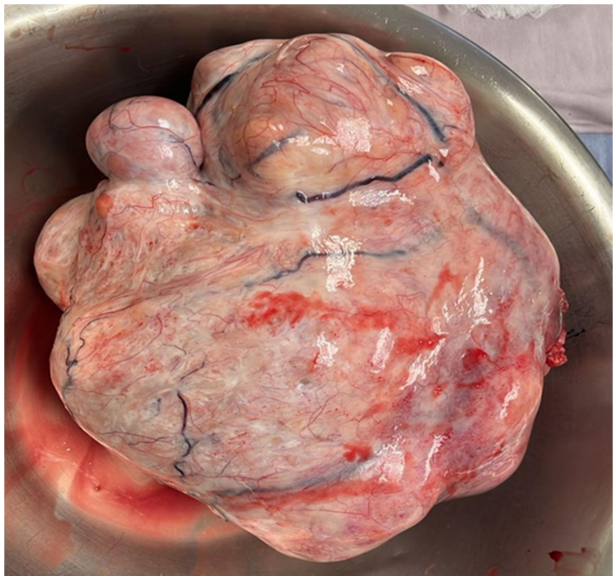

Figure 4. Macroscopic sample of ovarian mass of 1,027 g.

Surgical removal of the ovarian mass through laparotomy or with minimally invasive techniques, such as laparoscopy, with an intraoperative tissue sample sent for frozen section histologic confirmation is curative.[20] In postmenopausal women, a total abdominal hysterectomy with bilateral salpingo-oophrectomy is recommended.[21] Removal of the tumor results in the resolution of ascites and pleural effusion and normalization of CA-125 in Meigs and pseudo-Meigs syndrome.[22]

Conclusion

In patients with an ovarian mass, pleural effusion, and ascites, a thorough investigation is warranted to exclude ovarian cancer. However, it is not necessarily caused by a malignant condition, as in this case it was associated with a benign condition such as Meigs syndrome. Therefore, it is important to understand this association, as it leads to an excellent prognosis with the removal of the mass.

Conflicts of interests

The authors have no conflicts of interests.

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