Meigs Syndrome Presenting as Bilateral Pleural Effusion in the Reproductive Age-group: A Rare Case Report

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Abstract

Introduction: Meigs syndrome constitutes about 1% of ovarian tumors with a characteristic triad of benign ovarian tumors, ascites, and pleural effusion that usually resolves after resection of the tumor. It commonly presents as a right-sided pleural effusion in postmenopausal women, but it may rarely present as a bilateral (B/L) pleural effusion.

Case presentation: A 35-year-old female presented with progressive dyspnea, chest pain, and dry cough for 2 months. The patient had a similar history in the past for which she was treated with antituberculosis drugs for 2 months on the basis of pleural fluid analysis. There is no resolution of pleural effusion even after 2 months of treatment. Examination revealed bilateral pleural effusion. Ultrasoundography (USG) diagnostic thoracentesis revealed B/L exudative pleural effusion with low adenosine deaminase (ADA). On further evaluation, an USG abdomen reported ascites and soft tissue lesion on the right adnexa. Contrast-enhanced computerized tomography (CECT) abdomen confirmed well-defined soft tissue lesion of right ovarian origin with elevated serum cancer antigen 125 (CA125) (273 µ/mL). The patient underwent a right salpingo-oophorectomy, and the sample was sent for histopathological examination (HPE). Histopathological examination is suggestive of right cellular ovarian fibroma with positive desmin and Wilms tumor gene 1 (WT1) immunohistochemistry (IHC) markers. The patient showed clinical improvement after the resection of the tumor. Postoperative follow-up after 2 weeks showed complete resolution of effusion on chest radiograph and USG.

Conclusion: Although the incidence of Meigs syndrome is rare in the reproductive age-group female, it should be considered as one of the differentials in persistent and recurrent pleural effusion. Meigs syndrome usually (85%) presents as unilateral (U/L) and mostly right-sided effusion. In rare instances it can present as B/L pleural effusion due to transudation of ascitic fluid. An elevated serum CA125 test does not always indicate malignancy.

Keywords: Adnexal mass, Ascites, Pleural effusion, Cancer antigen 125, Case report, Diagnostic thoracentesis, Meigs syndrome, Reproductive age-group women.

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ABBREVIATIONS USED IN THIS ARTICLE

ADA = Adenosine deaminase; B/L = Bilateral; CA125 = Cancer antigen 125; CBNAAT = Cartridge-based nucleic acid amplification test; CECT = Contrast-enhanced computerized tomography; CT = Computed tomography; CXR = Chest X-ray; GHCCD = Government Hospital for Chest and Communicable Diseases; HPE = Histopathological examination; IHC = Immunohistochemistry; IL-1 = Interleukin 1; TNF-α = tumor necrosis factor-alpha; U/L = Unilateral; USG = Ultrasoundography; WT1 = Wilms tumor gene 1.

Introduction

Ovarian tumors considered as sixth most common cancer in women globally. Meigs syndrome is defined as the presence of a triad of pleural effusion, ascites, and benign ovarian tumor which usually resolves completely after resection of the tumor. Meigs syndrome occurs in 1% of ovarian tumors—thecoma, fibroma, Brenner tumor, or granulosa cell tumor. Ovarian fibroma is most commonly associated with Meigs syndrome. It is most common in postmenopausal women, and its incidence peaks in the seventh decade and is rare in reproductive age. Elevated cancer antigen 125 (CA125) levels with an adnexal mass, pleural effusion, and ascites in postmenopausal women are highly suggestive of malignancy.

Some patients with these findings may have a benign condition such as Meigs syndrome. After the removal of the tumor, this will disappear.

Case Presentation

A 35-year-old reproductive age-group women presented with complaints of progressive dyspnea, dry cough, and chest pain for 2 months to the Department of Pulmonary Medicine, Government
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Hospital for Chest and Communicable Diseases (GHCCD), Guntur, Andhra Pradesh, India. On the detailed evaluation, she revealed a history of weight loss. The patient had a similar history in the past for which she was treated with antituberculosis drugs for 2 months on the basis of pleural fluid analysis. There is no resolution of pleural effusion even after 2 months of treatment. On percussion, dull note is present over the lower half of the right and left hemi thorax. On auscultation, there were absent breath sounds on bilateral (B/L) infra-Scapular and infra-Axillary areas.

Chest radiograph showed B/L moderate pleural effusion (right > left) (Fig. 1). Ultrasonography (USG) diagnostic thoracentesis was done and biochemical analysis of fluid examination revealed a B/L exudative picture with right-side protein, 4.5 gm/L, and sugar, 98, with adenosine deaminase (ADA), 11; and left-side protein, 4.73 gm/L, and sugar, 143, with ADA-10.8. Cytological analysis of pleural fluid showed leukocytosis with 88% of leucocytes and 12% of polymorphonuclear cells and negative for malignant cells. Mycobacterium tuberculosis was not detected on both pleural fluid and sputum cartridge-based nucleic acid amplification test (CBNAAT). Table 1 shows pleural fluid analysis. Her viral markers such as HIV, hepatitis B surface antigen (HBsAg), and hepatitis C virus (HCV) were negative. Her complete blood picture, Hb, 11.8 gm; liver function tests; and renal function tests were within normal limits. Elevated serum CA125 was 273 µ/mL (normal, 35 µ/mL). The two-dimensional echocardiogram (2D echo) was normal. The patient was tachypneic with a saturation of 89% on room air, so we placed an intercoastal chest drain (ICD) on the right side.

Ultrasonography abdomen and chest reported moderate ascites and a large well defined heterogeneous hypo-echoic lesion of size 10 cm × 8.5 cm noted in the right adnexa with tiny internal cystic areas and moderate B/L pleural effusion. The CT chest showed B/L pleural effusion (Fig. 2). The contrast-enhanced computerized tomography (CECT) abdomen confirmed the

<table>
<thead>
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<th>Table 1: Pleural fluid analysis</th>
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<tr>
<td>Pleural fluid analysis</td>
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<tr>
<td>Protein</td>
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<tr>
<td>Sugar</td>
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<tr>
<td>TC</td>
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<td>ADA</td>
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<tr>
<td>Cytology</td>
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</tbody>
</table>

Figs 1A and B: (A) Chest radiograph (PA) view showing B/L pleural effusion (right > left); (B) Chest radiograph showing B/L pleural effusion with right-side ICD in situ

Figs 2A and B: Computed tomography (CT) chest (axial view) showing B/L pleural effusion
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The presence of abdominal pelvic mass of size 13.8 cm × 13 cm × 9.8 cm (Fig. 3) with no enhancement and irregular borders on contrast. The patient got referred to the Department of Gynecology for further evaluation and management. On follow-up at the Department of Gynecology, a right salpingo-oophorectomy was done on the patient and the sample was sent for histopathological examination (HPE). The HPE results suggested the right cellular ovarian fibroma (Fig. 4). The patient showed clinical improvement after resection of the tumor. Postoperative follow-up after 2 weeks showed complete resolution of effusion in the chest and abdomen (Fig. 5). Furthermore, CA125 decreased to 34 µ/mL during postoperative follow-up.

Discussion

Meigs syndrome was first reported in 1934, is defined as the triad of benign ovarian tumor, ascites, and pleural effusion that resolves after the removal of the tumor. It usually presents in postmenopausal women. About 2–5% of surgically removed ovarian tumors are fibromas. In 1% of ovarian tumors, Meigs syndrome is observed. In large tumors, pleural effusion is present in 1% and ascites in 10–15%. Low-grade malignant ovarian tumors with pleural effusion and ascites may sometimes be accepted as Meigs syndrome if the effusion disappears after surgical removal of the ovarian tumor. Pseudo-Meigs include ascites, pleural effusion, and an ovarian mass other than fibroma such as teratoma, mucinous cystadenoma, and metastatic malignancies of the ovary.

Meigs syndrome is common in postmenopausal women, however, our case was of the reproductive age-group female which is rare. It usually presents as right-sided pleural effusion, our patient presented with B/L pleural effusion, similar to Pr H Benjelloun et al., which is uncommon. Table 2 lists the cases of Meigs syndrome.

Patients usually present with shortness of breath, weight gain or weight loss, fatigue, nonproductive cough, increased abdominal girth, bloating, and menstrual irregularity in premenopausal women. Pleural fluid analysis ruled out tuberculosis, malignancy, and systemic causes of B/L pleural effusion, further we evaluated the patient with USG abdomen which revealed moderate ascites and a large well-defined heterogeneous hypoechoic lesion in the right adnexa. The CECT abdomen results confirmed the presence of abdominal pelvic mass with no enhancement and irregular borders on contrast. Her serum CA125 was 273 µ/mL. We referred the patient to the gynecology department for further management after stabilization.

Ascites are associated with large tumors and occur due to generalized secretion by the tumor itself. Exudative fluid is due to stimulation by the peritoneum’s mesothelial cells by interleukin 1 (IL-1), tumor necrosis factor-alpha (TNF-α), IL-8 vascular endothelial growth factor, and IL-6. The amount of pleural effusion developed is independent of the amount of ascites. The pleural effusion arises from the transdiaphragmatic transfer of ascitic fluid by lymphatic vessels, which are more on the right side. Rapid recurrence noted even after repeated thoracentesis.

One of the most common differential diagnoses for exudative pleural effusion is tuberculosis followed by parapneumonic effusions and malignancy. In tuberculosis-endemic countries such as India, Meigs syndrome is misdiagnosed as tuberculosis. It may be misdiagnosed as malignant effusion which leads to improper treatment. Furthermore, CA125 can be present in adult tissues such as ovaries, endometrium, endocervix, and fallopian tubes.
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Table 2: Cases of Meigs syndrome

<table>
<thead>
<tr>
<th>S. No.</th>
<th>Study</th>
<th>Age of presentation (years)</th>
<th>Laterality of pleural effusion</th>
<th>Histopathological diagnosis</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Miyu Tanaka et al.</td>
<td>54</td>
<td>Unilateral (U/L); right</td>
<td>Fibroma</td>
</tr>
<tr>
<td>2</td>
<td>Mongkol Benjapibal et al.</td>
<td>56</td>
<td>U/L; right</td>
<td>Fibroma</td>
</tr>
<tr>
<td>3</td>
<td>Javier Navarro–Esteva et al.</td>
<td>59</td>
<td>U/L; right</td>
<td>Fibroma</td>
</tr>
<tr>
<td>4</td>
<td>Javier Navarro–Esteva et al.</td>
<td>48</td>
<td>U/L; right</td>
<td>Fibroma</td>
</tr>
<tr>
<td>5</td>
<td>Divya Khanduja and Kajal NC</td>
<td>50</td>
<td>U/L; right</td>
<td>Fibroma</td>
</tr>
<tr>
<td>6</td>
<td>Hung–Wen Chen et al.</td>
<td>58</td>
<td>U/L; right</td>
<td>Fibroma</td>
</tr>
<tr>
<td>7</td>
<td>Cathy Monteith et al.</td>
<td>33</td>
<td>U/L; left</td>
<td>Leiomyoma</td>
</tr>
<tr>
<td>8</td>
<td>Aziz Slauoi et al.</td>
<td>69</td>
<td>U/L; right</td>
<td>Fibroma</td>
</tr>
<tr>
<td>9</td>
<td>Dipesh Upreti et al.</td>
<td>61</td>
<td>U/L; right</td>
<td>Fibroma</td>
</tr>
<tr>
<td>10</td>
<td>Guglielmo Stabile et al.</td>
<td>62</td>
<td>U/L; left</td>
<td>Fibrothecoma</td>
</tr>
<tr>
<td>11</td>
<td>Pr H Benjelloun et al.</td>
<td>51</td>
<td>B/L</td>
<td>Fibrothecoma</td>
</tr>
<tr>
<td>12</td>
<td>Wan-Chen Tsai et al.</td>
<td>84</td>
<td>U/L; right</td>
<td>Fibroma</td>
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</tbody>
</table>

It may also be present in mesothelial cells of the pericardium, pleura, and peritoneum. Low levels of serum CA125 can normally be present in the body. It is elevated during pregnancy or menstruation and in benign conditions such as cirrhosis or peritonitis or endometriosis. Above 35 µ/mL of CA125 is 60% for ovarian tumors and other malignancies are 84%. 

Conclusion

Though the incidence of Meigs syndrome is rare in the reproductive age-group female, it should be considered as one of the differentials in persistent and recurrent pleural effusion. Meigs syndrome usually (85%) presents as U/L and mostly right-sided effusion. In rare instances, it can present as B/L pleural effusion due to transduction of ascitic fluid. Elevated serum CA125 does not always indicate malignancy.

References

