Case Report

A Case of Synchronous Primary Corpus and Ovarian Cancer with Pseudo-Meigs Syndrome: Utilization of a Diagnostic Laparoscopy for the Accurate Diagnosis

Shimpei Shitanaka, Koji Yamanoi*, Jumpei Ogura, Tsutomu Ohara, Yoshihide Inayama, Takahiro Hirayama, Mie Sakai, Haruka Suzuki, Koji Yasumoto, Koh Suginami

Department of Obstetrics and Gynecology, Toyooka Public Hospital, Toyooka, Japan

Abstract

We report a case of synchronous primary corpus and ovarian cancer (SPC) with massive ascites due to Pseudo-Meigs syndrome (PMS). A 48-year-old woman presented with complaints of abnormal genital bleeding and abdominal discomfort. Massive ascites and tumors in the endometrium and right ovary were detected. Although imaging tests showed no evidence of dissemination, and ascites cytology was negative, we performed a diagnostic laparoscopy to exclude the possibility of microdissemination because pathological findings of the corpus tumor were suggested to be so-called Type-2 endometrial cancer. Laparoscopy clearly confirmed no dissemination in the peritoneum. We ultimately diagnosed this patient with SPC with massive nonmalignant ascites due to PMS and performed an appropriate treatment. This report is the first case of SPC that developed PMS.

Keywords: Diagnostic laparoscopy, Pseudo-Meigs syndrome, synchronous primary corpus and ovarian cancer

Introduction

Malignant ascites frequently develops when a malignant tumor is present in the pelvis. However, the tumors cause nonmalignant ascites in some cases. This condition is known as Pseudo-Meigs syndrome (PMS).

Meigs syndrome is the triad of ascites, pleural effusion, and benign fibrous ovarian tumors, including ovarian fibroma, fibrothecoma, and Brenner tumor. When tumors are not fibromas, the condition is defined as PMS.[1] Notably, ascites resolves after resection of the tumor.

Synchronous primary endometrial and ovarian cancer (SPC) is frequently encountered clinically. We previously reported several characteristics of SPC,[2] but, to date, there are no reports referring to PMS and SPC.

We report a case of SPC with massive nonmalignant ascites due to PMS. A diagnostic laparoscopy markedly helped us make an accurate diagnosis and choose an appropriate treatment.

Case Report

A 48-year-old woman (gravida 4, para 2) visited us with complaints of abnormal genital bleeding and abdominal discomfort for the past 3 months. Her menstrual cycle was regularly for approximately 28 days.

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Address for correspondence: Dr. Koji Yamanoi, Department of Dermatology, Cutaneous Biology Research Center, Massachusetts General Hospital, Boston, MA, USA.
E-mail: kojiymni@kuhp.kyoto-u.ac.jp

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We found massive ascites and tumors in the endometrium and right ovary on ultrasound examination. We performed a hysteroscopy and biopsied the corpus tumor. A high columnar cell with high-grade atypia proliferated in the tubular and papillary in corpus tumors, which suggests the so-called Type-2 endometrial cancer (endometrial cancer; Grade 3 or serous adenocarcinoma) [Figure 1a].

We performed magnetic resonance imaging (MRI) and computed tomography (CT).

The tumor in the endometrium showed low-signal intensity (SI) in T2-weighted images (T2WIs), with a contrast effect in the T1WI. Tumor invasion did not appear to include more than half of the uterus myometrium. The right ovarian tumor was 10 cm in the diameter and consisted of irregular solid and cystic parts. The solid part showed a high SI in the T2WI, T1WI, and diffusion-weighted images with contrast effect [Figure 1b and c]. CT images revealed the presence of massive ascites. There was no evidence of peritoneal dissemination or any distal metastasis [Figure 1d].

Tumor markers carcinoembryonic antigen, carbohydrate antigen 125 (CA125), and CA19-9 were elevated markedly compared to the normal ranges (15.6 ng/mL, 240 U/mL, and 2745.9 U/mL, respectively). We performed abdominocentesis followed by cytology, and tumor cells were not detected in the ascites.

Based on these results, PMS was suspected as the reason of her massive ascites. However, a literature survey revealed no reports about PMS and corpus cancer or SPC. We considered microdissemination as the reason for her massive ascites because corpus tumors are considered a more malignant phenotype. To investigate the state of the abdominal cavity precisely, we performed a diagnostic laparoscopy.

We performed single-port laparoscopic surgery using the transumbilical approach with a 3-cm midline incision. A large volume of yellow ascites was present in the abdominal space. Up to 6600 ml of ascites was aspirated at the beginning of surgery [Figure 1e]. We performed cytology at different time points during aspiration, and the results were all negative. The tumor in the right ovary had enlarged to fist size and ruptured spontaneously [Figure 1f]. The left adnexa and serosa of the uterus were intact [Figure 1g]. We observed the abdominal space carefully and there was no peritoneal dissemination [Figure 1f-h]. The right adnexa was resected laparoscopically and collected using a laparoscopic bag. We absorbed all ascites at the end of laparoscopy [Figure 1h]. Endometrial curettage was performed simultaneously. The operation time was 2 h and 37 min, and the intraoperative blood loss, including ascites, was 1060 ml.

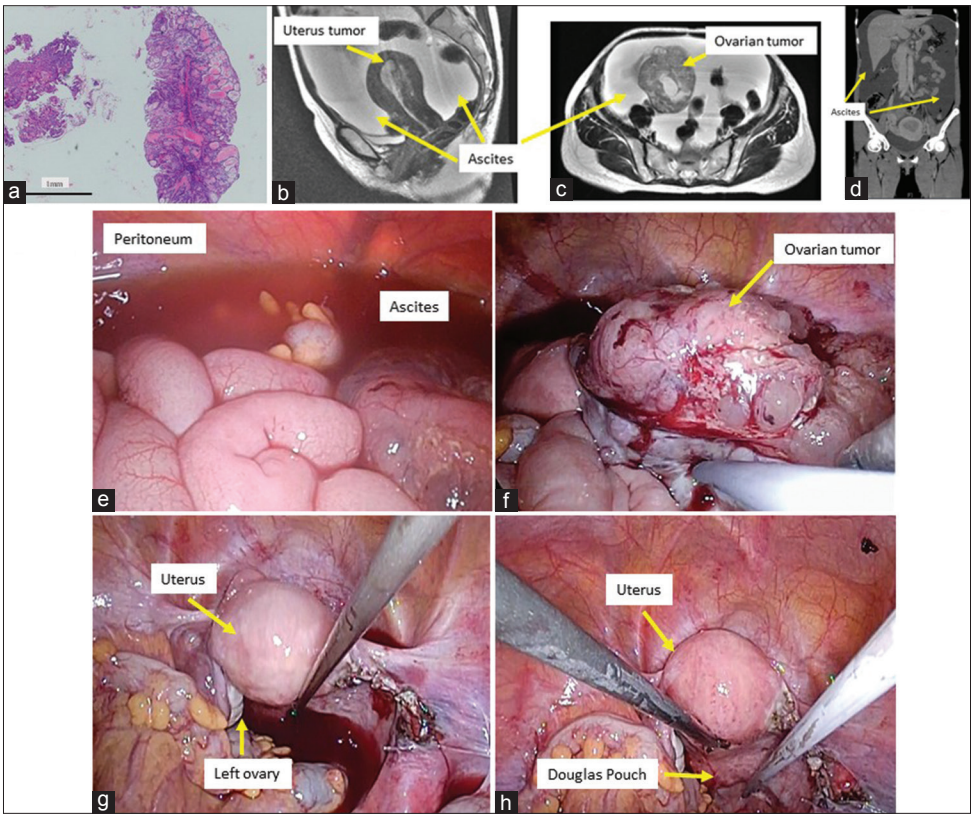

Figure 1: Preoperative findings of pathology, imaging tests and intra-operative views. (a) Corpus tumor, H and E, ×40. (b-d) Imaging tests findings; magnetic resonance imaging (T2-weighted images) (b and c) and computed tomography (d). (e-h) Intraoperative findings in the diagnostic laparoscopy

The patient had an uneventful postoperative course, and she was discharged on the 3rd postoperative day. We performed contrast-enhanced CT on the 8th postoperative day and found that ascites reappeared. However, the volume was low compared to prior to surgery [Figure 2a and b].

Pathologically, a high columnar cell with high-grade atypia proliferated in the tubular right ovarian tumor with mucin production. There was no fibroma and stromal edema. Immunohistochemistry staining was strongly positive for p53, Vimentin, the progesterone receptor, and the estrogen receptor (ER). A high columnar cell with high-grade atypia proliferated in corpus tumors in the tubular and papillary region. The tissue was negative for the ER and strongly positive for p53 [Figure 2c and d].

We diagnosed this case as SPC: cStage IA corpus cancer (endometrial adenocarcinoma, Grade 3) and cStage IC2 right ovarian cancer (high-grade endometrioid adenocarcinoma). The massive ascites was due to PMS.

Primary cytoreductive surgery (abdominal simple total hysterectomy, left sapling-oophorectomy, and partial omentectomy) with retroperitoneum lymph node dissection was performed on the 17th postlaparoscopic surgery day. Some yellow clear ascites were present at the beginning of surgery, and its cytology was negative. Little adhesion existed, and there was no peritoneal dissemination. The pathological findings were similar to previous findings [Supplementary Figure 1]. We ultimately diagnosed this case as SPC: stage IA corpus cancer (endometrial adenocarcinoma, Grade 3) and Stage IC2 ovarian cancer of the right (high-grade endometrioid adenocarcinoma).

The patient had an uneventful postoperative course, and she was discharged on the 17th postoperative day. Ascites was absent after surgery. Six cycles of combination chemotherapy of paclitaxel and carboplatin were administered after surgery. The patient is currently undergoing regular follow-up without any evidence of recurrence.

Discussion

The present report described a case of SPC with massive ascites due to PMS. To the best of our knowledge, this report is the first case of SPC with PMS.

We previously reported some characteristic clinic pathological features of SPC, including the age at diagnosis, better prognosis, and a high frequency of endometriosis and thrombosis.[2] The present case was diagnosed at the relatively young age of 46 years, and her menstruation was regular, which are compatible with these features. Several malignant diseases that cause PMS were reported, including ovarian cancer, metastatic ovarian cancer, and large leiomyoma of the uterus.[3-5] However, there are no reports of PMS with SPC and/or primary corpus cancer. Therefore, PMS is likely not

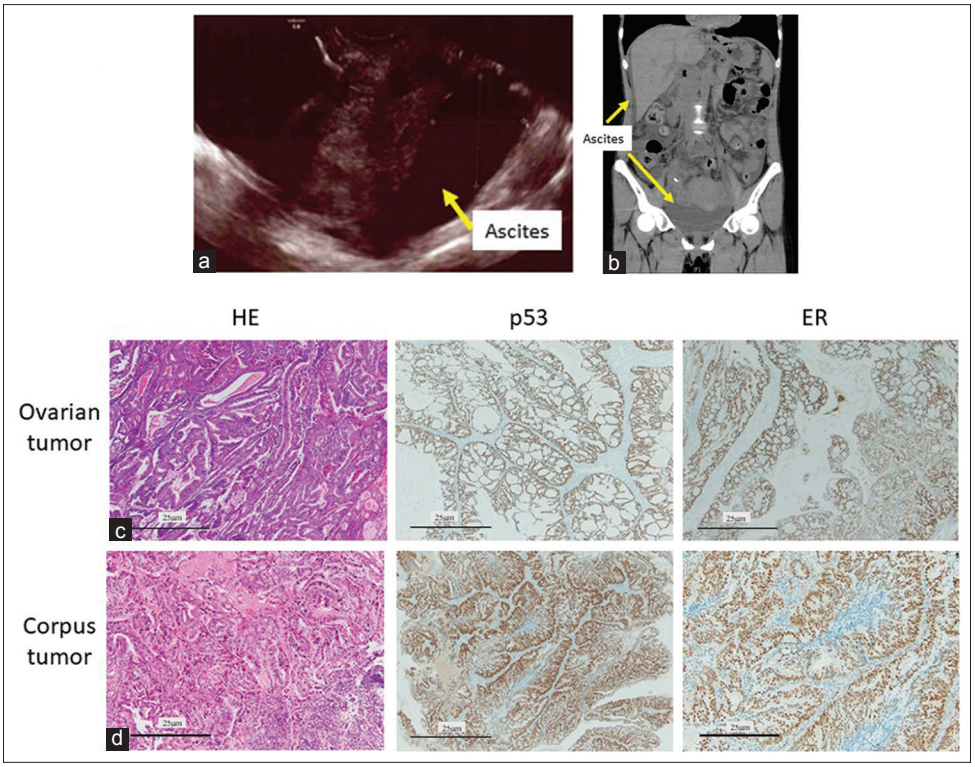

Figure 2: Findings of imaging tests after laparoscopy and pathological findings of tumor. (a and b) Imaging test findings; ultrasound examination (a) and computed tomography (b). (c and d) Pathological findings of tumor of ovary (c) and corpus (d); ×40. Left: HE, Middle: p53, Right: ER

characteristic of SPC or corpus cancer. The reason that PMS developed in this case is not clear.

Several hypotheses about the mechanism of ascites accumulation in PMS were proposed. Some studies suggest that ascites results from stromal tumor edema.[6] Some studies also suggest that it is linked to inflammatory cytokines and/or growth factor release, including vascular endothelial growth factor and interleukin-6.[7]

Removal of the right ovarian tumor largely decreased the amount of ascites in the present case, so massive ascites might be derived from the cyst fluid of the ovarian tumor. However, ascites did not completely disappear. We hypothesize that some inflammatory cytokines and/or growth factors played an important role in the production of ascites in this case because the corpus cancer was not very large, and it was not exposed to the peritoneal cavity. Further investigations are needed to elucidate the detailed mechanisms of PMS.

The absence of cancer cells in the ascites, and peritoneal dissemination must be confirmed for a diagnosis of PMS. Imaging tests, such as CT, MRI, and cytology, are common methods, but some limitations exist.

de Bree *et al*. reported that the detection of individual peritoneal disseminations varied from 9.1% to 24.3% for tumor sizes <1 cm.[8] Bando *et al*. have reported that the cytological positivity rate was approximately 50% for patients who have dissemination in the peritoneum cavity.[9] These results suggest that dissemination cannot be accurately predicted using only imaging tests and/or cytology.

The diagnostic laparoscopy is useful when it is difficult to make an accurate diagnosis. We reported previously that a diagnostic laparoscopy was useful in a case of gliomatosis peritonitis followed by treatment of the immature teratoma.[10] Laparoscopic surgery reduces intraoperative blood loss, postoperative pain, and the length of hospital stay compared to laparotomy. We performed primary cytoreductive surgery on 17th postoperative day in the present case. If an accurate diagnosis is difficult to obtain, close observation with laparoscopy may be considered.

Conclusion

We report a case of SPC with massive ascites due to PMS. A diagnostic laparoscopy markedly helped us obtain an accurate diagnosis of PMS and choose an appropriate treatment. The use of only imaging tests and/or cytology to assess status may occasionally be insufficient. The diagnostic laparoscopy should be considered in these cases to obtain an accurate diagnosis.

Ethical statement

This study is approved by IRB of Toyooka Public Hospital, approval no. 199 obtained on July 31th, 2019.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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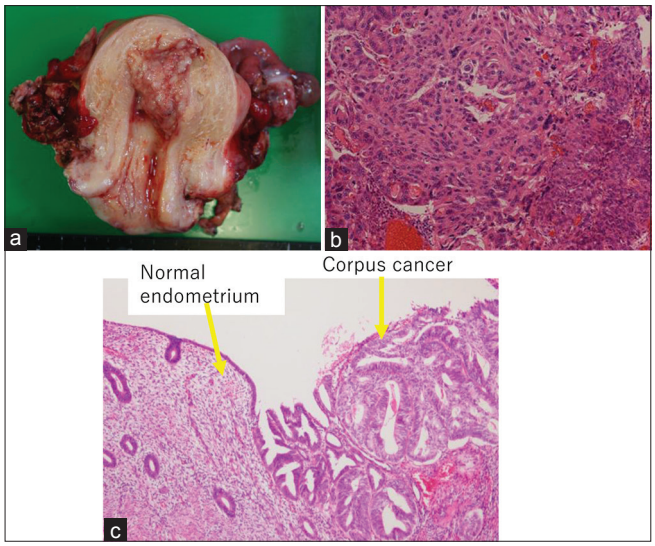

Supplementary Figure1: Macroscopic and microscopic findings of uterus tumor; primary cytoreductive surgery. (a) Macroscopic finding of uterus. Myometrial invasion of the tumor was less than half. (b) Uterus tumor, x40 magnification of hematoxylin and eosin (HE). (C) Uterus tumor, x40 magnification of hematoxylin and eosin (HE)