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CASE REPORT

Benign Multicystic Peritoneal Mesothelioma Discovered during Gynecologic Laparoscopic Surgery after Chemotherapy: A case report

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ABSTRACT

Benign multicystic peritoneal mesothelioma (BMPM) is an extremely rare intraperitoneal tumor that has been associated with endometriosis, pelvic inflammatory disease, and previous abdominal surgery. BMPM is a benign disease with an unknown etiology and clinical problems. The woman with no childbearing history developed intravascular large cell lymphoma at the age of 42 years, was treated with rituximab, cyclophosphamide, doxorubicin, vincristine, and prednisolone (R-CHOP). After chemotherapy, the patient developed ovarian insufficiency, and gradually increased the left ovarian cyst of 7 cm in diameter. We planned laparoscopic bilateral adnexectomy to prevent left ovarian torsion and to search for pathology. After surgery, these cysts were pathologically diagnosed as BMPM. Problems of BMPM are high rate of recurrence, impairing quality of life and fertility due to causing in young age. Chemotherapy would be one of risk factor causing BMPM, since there are many reports of BMPM occurring in young women, gynecologists should take fertility into consideration when treating these patients.

Keywords: benign multicystic peritoneal mesothelioma, chemotherapy, infertility.

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Introduction

Benign multicystic peritoneal mesothelioma (BMPM) is an extremely rare intraperitoneal tumor that has been associated with endometriosis, pelvic inflammatory disease, and previous abdominal surgery[1]. BMPM is a benign disease with an unknown etiology and clinical problems such as a high recurrence rate, the possibility of malignant transformation, and lymph node metastasis[2]. It is difficult to diagnose and is often found incidentally during surgery[3], and there have been almost no reports in the field of gynecology. In this report, we describe a rare case of BMPM that was incidentally discovered during laparoscopic surgery after chemotherapy.

Case

A 46-year-old Japanese woman with no childbearing history developed intravascular large cell lymphoma at the age of 42 years, and was treated with rituximab, cyclophosphamide, doxorubicin, vincristine, and prednisolone (R-CHOP) and subsequently achieved remission. After chemotherapy, the patient developed ovarian insufficiency and was referred to the obstetrics and gynecology department where hormone replacement therapy was started. During the course of treatment and observation, a gradually increasing left ovarian cyst of 7 cm in diameter and an increasing serous cyst of 7 cm in diameter just below the fascia in the lower abdomen were noted (Fig. 1). Although the patient had no clinical symptoms, laparoscopic bilateral adnexectomy was planned to prevent left ovarian torsion and to search for pathology.

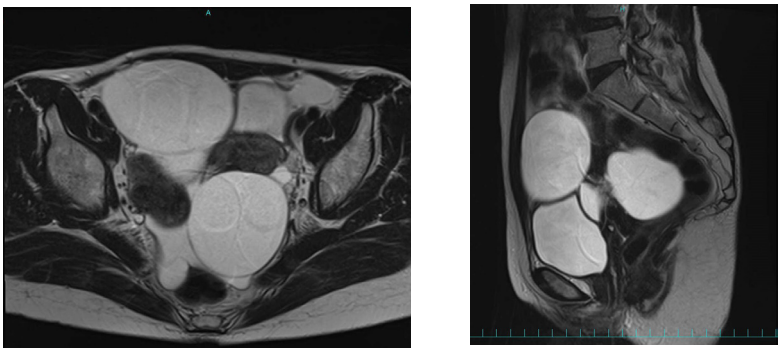

Fig. 1. MRI findings left ovarian cyst of 7 cm in diameter and a serous cyst of 7 cm in diameter just below the fascia in the lower abdomen.

Prior to laparoscopic surgery, a 7 cm serous cyst was found just below the fascia, which may have interfered with the laparoscopic approach, so the department of radiology was requested to aspirate the fluid from the cyst using interventional radiology. The contents were later found to have the same properties as a BMPM based on cytologic exploration. Laparoscopic surgery was performed in parallel approach, and intraperitoneal findings included a left ovarian cyst as well as colorless to pale yellow serous polycystic lesions scattered throughout the abdominal cavity. The cysts were extensively spread throughout the pelvic cavity, including the ovarian surface, fallopian tubes, peritoneum, omentum, and gastrointestinal tract (Fig. 2). The multiple cysts in the pelvis made it difficult to differentiate BMPM on preoperative imaging. After intraperitoneal observation, laparoscopic bilateral adnexectomy was performed as well as several biopsies of the polycystic lesions to complete the procedure. The pathological diagnosis of BMPM was made based on the positive

results of immunostaining for calretinin, podoplani, and Wilms' tumour protein (WT-1) and negative results for cluster of differentiation 31 (CD31) (Fig. 3). The patient was discharged on the fourth postoperative day in good general condition. The BMPM was referred to a gastrointestinal surgeon at another hospital, and the subfascial cyst was aspirated according to abdominal symptoms.

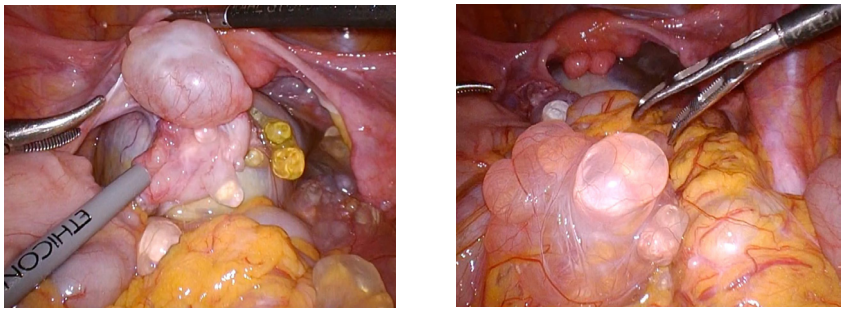

Fig. 2. Laparoscopic finding: the cysts were extensively spread throughout the pelvic cavity, including the ovarian surface, fallopian tubes, peritoneum, omentum, and gastrointestinal tract.

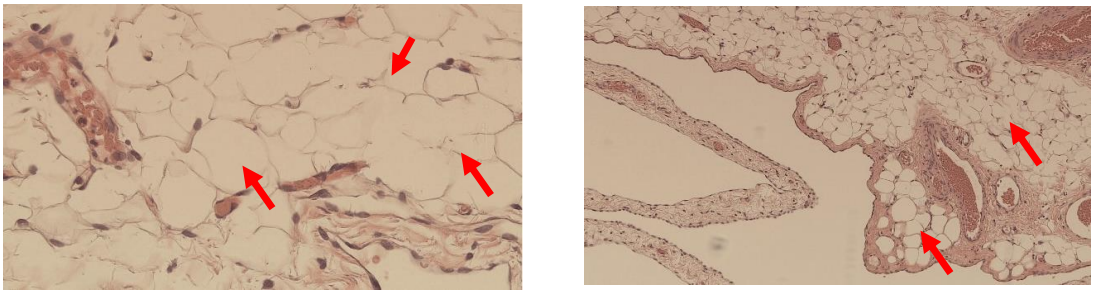

Fig. 3. The pathological diagnosis of benign multicystic peritoneal mesothelioma based on the positive results of immunostaining for calretinin, podoplani, and WT-1 and negative results for CD31.

Discussion

BMPM was first described by Menemeyer and Smith in 1979[4]. It is often difficult to diagnose preoperatively because it may be discovered incidentally during surgery. MRI is the best imaging technique for BMPM but definitive diagnosis relies primarily on histopathological examination and immunohistochemistry, especially, the D2-40 stain can strongly promote a diagnosis of BMPM[1].

While surgical removal of the tumor en bloc is recommended as the primary treatment strategy[1], Alpár György reported a high recurrence rate of 50% for BMPM[5]. Thus, recently BMPM is considered as a borderline malignant neoplasm because of high recurrence rate[1]. Eran Nizri reported that, in addition to surgery, hyperthermic intraperitoneal chemotherapy (HIPEC) is effective[6]. Pathologic diagnosis is difficult[7], and differentials include lymphangioma, peritoneal pseudomyxoma, endometriosis, ovarian cystadenoma or cystadenocarcinoma, cystic teratoma, and other large mesenteric cysts, and differentiation from malignant mesothelioma is most important issue[1].

Risk factors for developing BMPM include being female, having endometriosis, leiomyoma, pelvic inflammatory disease, and a history of abdominal

surgery of either sex[3]. Genetic association has been suggested, but no clear responsible gene has been identified. Of note, malignant transformation is a possibility, but is reported to be infrequent and is often observed in the absence of symptoms. The present patient had no pelvic inflammatory disease, history of surgery or medical history of endometriosis, but had received chemotherapy several years before onset of disease. In other reports, BMPM has been reported incidentally after referral to obstetrics and gynecology after early menopause due to postoperative adjuvant chemotherapy for breast cancer, suggesting that chemotherapy may be involved in the development of BMPM[2]. Several physicians believe that reactive etiology is the etiology, and the presence or absence of chemotherapy, especially in young women, may be a risk factor for BMPM[8].

There are few reports in the field of gynecology that point to an association between BMPM and ovarian tumors. Thus, gynecologists should be aware of the presence of BMPM when they discover multiple cystic lesions in the abdominal cavity preoperatively or intraoperatively, taking risk factors into consideration. If clinical symptoms develop, surgical treatment should be considered according to the patient's quality of life. In principle, complete surgical resection is recommended, but such as the present case, where there are no symptoms and complete removal is difficult because the tumor has spread throughout the abdominal cavity, partial removal may be acceptable according to pathological exploration purposes and reducing symptoms. In addition, malignant transformation rate is extremely low. The patient's treatment history, including chemotherapy, should be taken into consideration. Since there are many reports of BMPM occurring in young women, gynecologists should take fertility into consideration when treating these patients. The frequency of BMPM has not been systematically reported, and the accumulation of future cases may help guide treatment.

Potential conflicts of interest

The authors declare no conflicts of interest.

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