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Case report

# Multicystic peritoneal mesothelioma after fertility-sparing surgery for an ovarian tumor of borderline malignancy: A case report

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ABSTRACT

We report a case of a 19-year-old woman with multicystic peritoneal mesothelioma (MCPM) who had previously undergone left adnexectomy due to an ovarian tumor of borderline malignancy at the age of 17 years. Follow-up imaging studies after adnexectomy revealed multiple cystic lesions of increasing size and number, suggesting recurrence of the tumor. Diagnostic laparoscopic surgery was performed, and the cystic lesion was pathologically determined to be MCPM. To our knowledge, this is the first report of MCPM diagnosed and successfully treated by laparoscopic surgery during the course of follow-up for an ovarian tumor. It is important to recognize that MCPM can occur in patients who have previously undergone abdominal surgery, and laparoscopic surgery is recommended for patients of reproductive age, because of the potential risk of infertility associated with extensive pelvic surgery.



## Introduction

Approximately 150 cases of multicystic peritoneal mesothelioma (MCPM) have been reported in the medical literature since it was first described by Mennemeyer and Smith in 1979.[1] According to currently available literature, MCPM most commonly occurs in the abdominopelvic cavity of women of reproductive age who have no history of asbestos exposure, but who do have a history of previous abdominopelvic operations, endometriosis, or pelvic inflammatory disease. Although considered by many to be benign,[2] this tumor has a high local recurrence rate and is usually found incidentally in asymptomatic patients. MCPM is usually found in the peritoneal cavity, intimately attached to surfaces of the intestine, omentum, spleen, liver, or retroperitoneal space, and is rarely found at extra-abdominal locations.[3] MCPM is most often found in women (female:male ratio, 5:1); however, there are reports of the disease diagnosed in men[4] and children as well.[5]

In this case, a cystic lesion was discovered by imaging modalities during routine follow-up imaging for an ovarian tumor of borderline malignancy. Diagnostic laparoscopic surgery was performed and showed that the cystic lesion was MCPM. To our knowledge, this is the first report of MCPM diagnosed and successfully treated by laparoscopic surgery during routine follow-up for an ovarian tumor of borderline malignancy. It is important to recognize that MCPM can occur in patients who have previously undergone abdominal surgery, and laparoscopic surgery is recommended for patients of reproductive age because of the potential risk of infertility associated with extensive pelvic surgery.[6]

## Case report

A 17-year-old nulliparous woman was referred to our hospital for treatment of an ovarian tumor. She had no history of major illnesses or surgery, and her family history was unremarkable. Her preoperative serum levels of carbohydrate antigen (CA)-125 and CA19-9 were 175.6 U/mL (range, 0–35 U/mL) and 176.2 U/mL (range, 0–37 U/mL), respectively. Magnetic resonance imaging (MRI) revealed a large, 20 cm, multicystic tumor in the abdominal cavity that did not have a solid component or show enhancement with gadolinium. Cytology analysis of the ascites fluid did not show the presence of cancer cells, and no malignant lesions, other than that in the left ovary, were detected. A preoperative diagnosis of an ovarian tumor was made, and laparotomy was performed. During surgery, the pathological diagnosis based on frozen section analysis

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of the tumor was mucinous cystadenoma, and only left adnexectomy was performed. Following surgery, the final pathological diagnosis was a mucinous cystic tumor with borderline malignancy, but as the patient wished to bear children, no further treatment was administered.

Seventeen months after surgery, routine follow-up with computed tomography revealed six cystic lesions, each of which was 1 cm in size, in the pelvis, but the patient's serum levels of CA-125 and CA19-9 were 15.5 U/mL and 9.4 U/mL, respectively (within normal limits). MRI indicated no solid or enhanced components in the cystic lesions, indicating no apparent malignancy. However, on close observation for an additional year, we noted an increasing amount of ascites fluid and at least eight cystic lesions, each of which was 2–3 cm in size, in the pelvis (Fig. 1). These lesions showed low to intermediate intensity on T1-weighted images, high intensity on T2-weighted images, and no enhancement on administration of gadolinium. Although serum levels of CA-125 and CA19-9 remained within normal limits, MRI findings suggested the possibility of recurrent disease. To confirm this finding, diagnostic laparoscopic surgery was performed, beginning with an evaluation of the abdominal cavity. Grape-like cystic structures developing in the cul-de-sac, on the pelvic sidewall on both sides, and on the lower portion of the greater omentum were identified (Figs. 2 and 3), and incisional biopsy was performed. No cystic lesions were detected in the upper abdomen. Cytopathologic examination of the abdominal fluid did not show the presence of any malignant cells, and the pathologic diagnosis of a frozen tissue section during surgery was peritoneal mesothelioma without malignancy. All recognizable cysts were removed, and partial omentectomy was performed to preserve fertility. The patient had an uneventful recovery and was discharged from the hospital.

Gross examination of the tissues obtained from the patient revealed a multicystic mass with no solid areas within the lesion. Individual cysts ranged from 1 cm to 5 cm in diameter and contained serous fluid. The walls of the cysts were semitransparent, < 1 mm thick, and lined with a single layer of flat cells that were immunopositive for calretinin (Fig. 4). No columnar, malignant cells containing mucin were detected. The patient is currently recurrence free 1 year following the laparoscopic surgery for MCPM, has regular menstruation, and is being regularly monitored for recurrence.

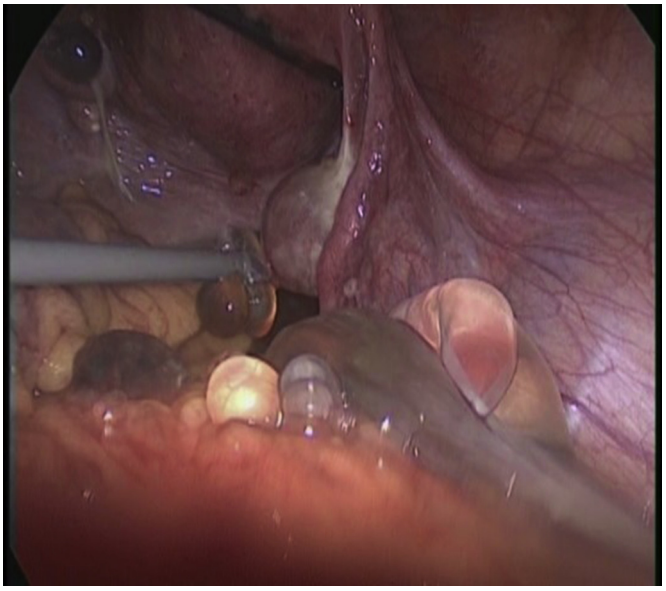

**Fig. 2.** Intraoperative findings of the pelvis revealed multiple cystic lesions.

## Discussion

MCPM is a rare, localized tumor arising from the epithelial and mesenchymal elements of the mesothelium and has most commonly been described in adult women of a median age of 36 years.[7] Most MCPMs are diagnosed incidentally. However, some patients present with a localized mass or with abdominal pain.[8] To our knowledge, this case report is the first to describe MCPM that was diagnosed and treated by laparoscopic surgery during the course of follow-up for a gynecologic malignancy.

The pathogenesis of MCPM is controversial. Some reports suggest that MCPM is neoplastic, whereas others point to a reactive process.[9,10] As most patients who develop MCPM have a previous history of intrapelvic surgery for endometriosis, uterine leiomyoma, or inflammation, development of this tumor has been suggested to be a peritoneal reaction to chronic irritation with mesothelial cell entrapment, reactive proliferation, and cyst formation.[8] Histologically, MCPM presents as cystic spaces lined by

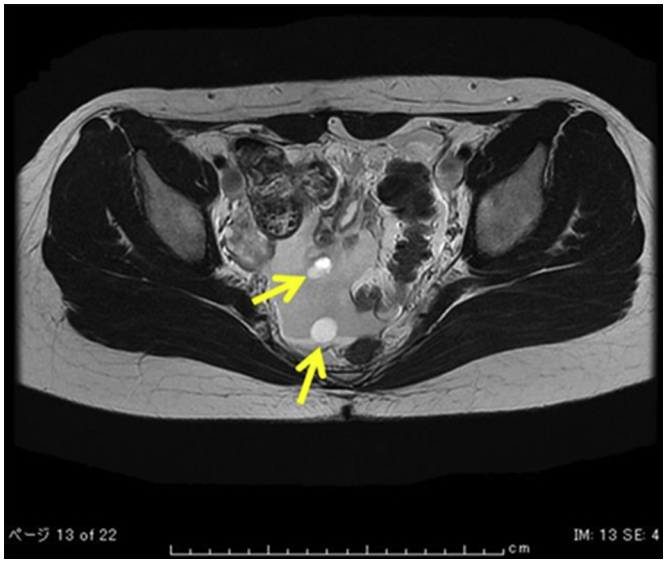


**Fig. 1.** Transverse magnetic resonance imaging of the pelvis shows ascites and cystic lesions (arrows) on a T2-weighted image.

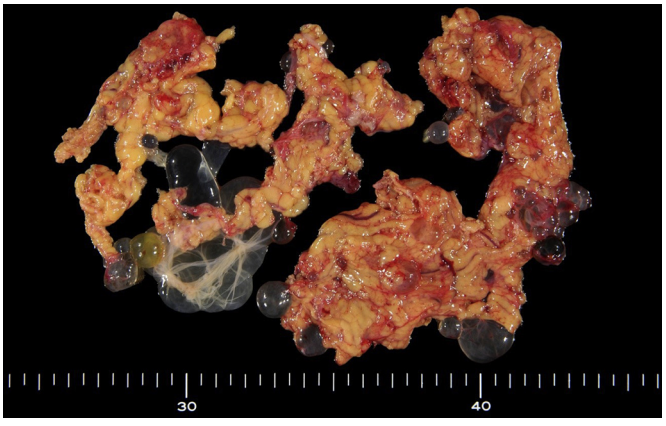


**Fig. 3.** The gross appearance of the omentectomy specimen following laparoscopic surgery is shown.

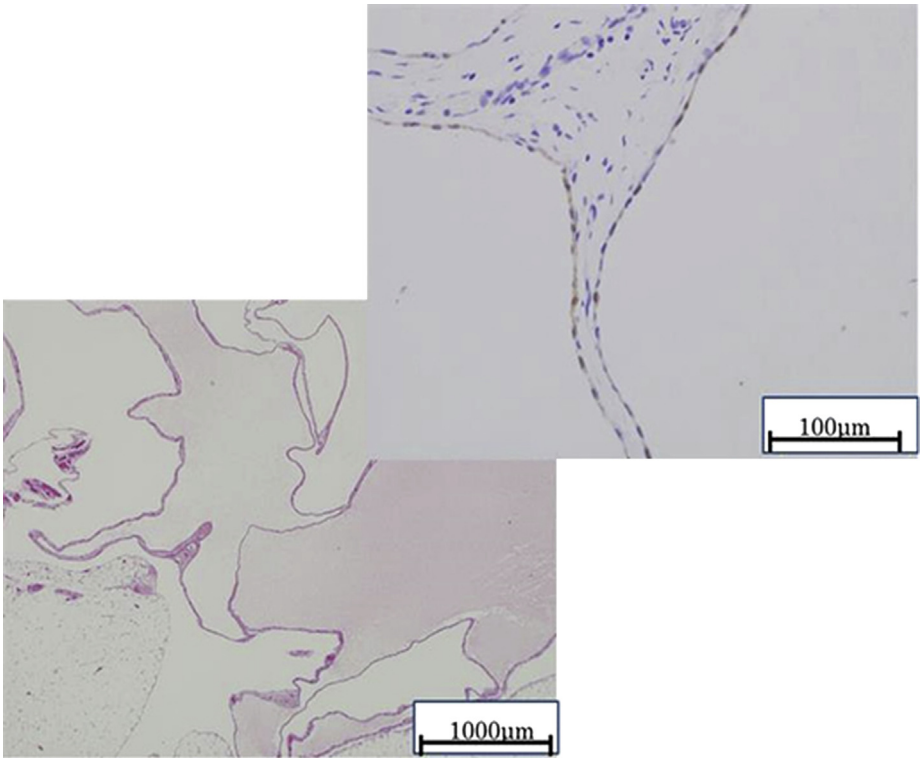


**Fig. 4.** The walls of the cysts are lined with a single layer of flat cells that are immunopositive for calretinin.

low cuboidal cells, with hobnail features and prominent vascularity. The primary differential diagnosis for multilocular cysts of the peritoneum is either cystic lymphangioma or MCPM. Whereas MCPM shows positive immunohistochemical reactivity for calretinin, lymphangioma is immunohistochemically negative for calretinin and shows positive immunohistochemical reactivity for D2-40. As most patients presenting with MCPM are women of reproductive age, female sex hormones are suggested to play a key role in the pathogenesis,[8] and this disease may develop in patients undergoing fertility-sparing surgery. When cystic lesions are detected during the course of follow-up, a careful diagnosis must be made, and less aggressive approaches for remediation should be favored, because of the risk of infertility associated with extensive pelvic surgery in women.[6] Therefore, laparoscopic surgery is effective and recommended for an accurate diagnosis.

MCPM commonly presents with an indolent clinical behavior; however, early recurrence requiring multiple surgical interventions,[6,10,11] transformation into truly malignant disease,[12] lymph node involvement,[13] and death[14] following MCPM diagnosis have been described. In this case report, we confirm the findings of the study by Baratti et al[15] by showing that MCPM is a locally aggressive and borderline malignant tumor capable of transition into an invasive and potentially lethal process, rather than a benign disease, and suggest that long term follow-up is necessary for proper management of this disease.

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