Case Report
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Multicystic benign mesothelioma of the pelvic peritoneum presenting as acute abdominal pain in a young woman

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Multicystic benign mesothelioma (MBM) of the peritoneum is a very rare condition. Since the first description of MBM in 1979, approximately 100 cases have been reported. This is a case report of MBM of the pelvic peritoneum presenting as acute abdominal pain in a young woman. Laparoscopy confirmed multiple grapelike clusters of cysts that originated in the peritoneum of the rectouterine pouch and histopathologic diagnosis was confirmed as MBM of the pelvic peritoneum. We hope to alert gynaecologists of the diagnostic and therapeutic approaches to MBM which can be accomplished by laparoscopy.

Keywords: Abdominal pain; Acute; Benign; Mesothelioma

Introduction

Multicystic benign mesothelioma (MBM) of the peritoneum is a very rare disease. Since the first description of MBM in 1979, approximately 100 cases have been reported. The preoperative diagnosis is difficult and covers the whole range of cystic and papillary lesions of the peritoneum and the ovaries [1,2]. This is a case report of MBM of the pelvic peritoneum presenting as acute abdominal pain in a young woman.

Case Report

A 28-year-old woman, gravida 2, para 0, abortion 2, had been experiencing progressive lower abdominal pain for almost 7 months. The pain worsened a week prior to her admittance and remained constant. She denied fever or chills. Her appetite, bowel and urinary function were normal and she had no recent weight loss. She experienced regular menstrual cycles and a long-standing history of dysmenorrhea and dyspareunia. Past medical history was unremarkable and there was no history of previous abdominal surgery or gynecologic disorders. Family history was not notable.

On physical examination, the abdomen was mildly rigid and moderately distended. She had a tender mass behind the uterus but the cervix and genital tract appeared normal. Complete blood screening profile, urinalysis and serum CA-125 assay were within normal limits. Ultrasonography and computed tomography of the pelvis revealed a 10 × 7 cm multicystic mass with irregular borders occupying the rectouterine pouch (Fig. 1).

We decided to perform laparoscopic surgery. Laparoscopic trochars were placed below the umbilicus and lower abdomen (×3, 12 mm, 5 mm, 5 mm). During the laparoscopy, we observed multiple grapelike clusters of cysts within the rectouterine pouch. The cysts appeared to originate from the peritoneum of the rectouterine pouch and attached to the uterus and left adnexa. Additional small cysts were found

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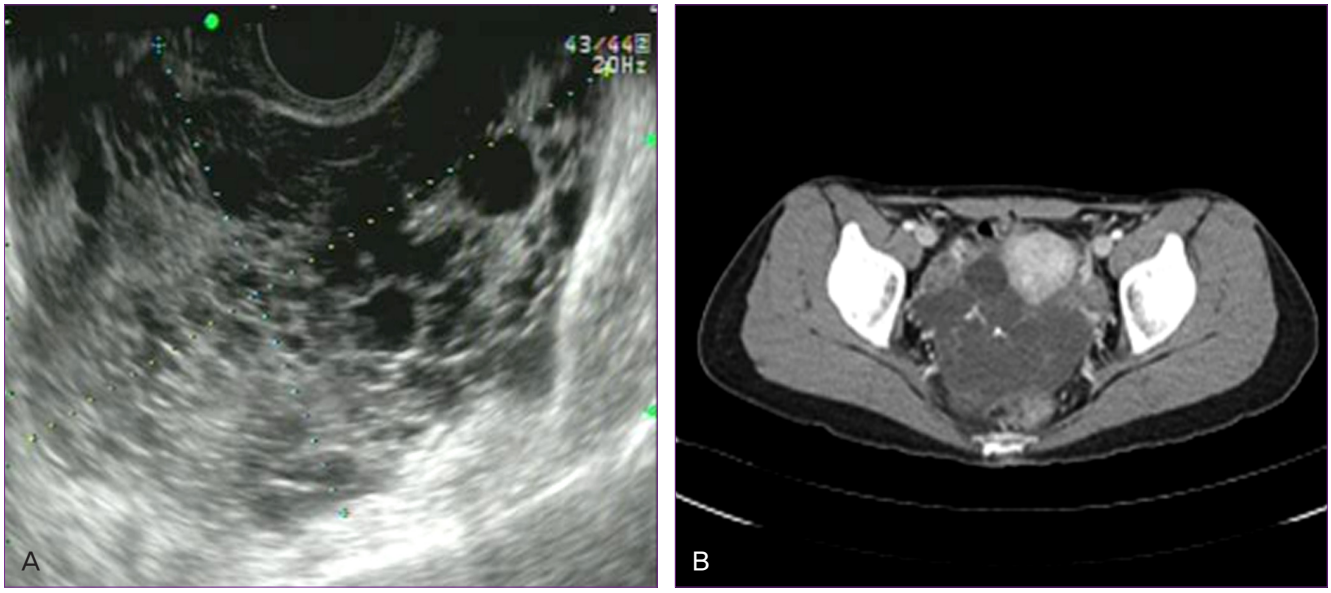

Fig. 1. (A) Large multilocular hypoechogenic cysts with irregular border on vaginal ultrasonography. (B) Contrast enhanced computed tomography with a hypodense, multicystic mass with contact to the uterus without infiltrative growth, ascites or lymph node involvement.

on the greater omentum and serosal surface of the right fallopian tube. Careful exploration of the whole abdomen, including appendix, liver and diaphragm, revealed no other abnormalities. Intraoperative frozen section histology of the multiple grapelike clusters of cysts were suspicious for serous type benign cysts. A decision was made to excise all macroscopically visible cysts. Excision of multiple clusters of cysts in the rectouterine pouch via laparotomy, resection of the peritoneum of the rectouterine pouch, partial greater omentectomy and electro-fulguration (argon plasma coagulator) of small cysts on the serosal surface of the right fallopian tube was performed.

Grossly, multiple cysts were present measuring up to 2 cm. Microscopically, the cysts were lined by flattened or cuboidal cells. The lining cells were reactive immunohistochemically for Pan-CK, calretinin, D2-40 and CK5/6 but negative for CD34 (Fig. 2). Histological examination of the excised specimens were consistent with a MBM of the pelvic peritoneum. The postoperative course was uneventful. The patient is well at 3-month follow-up with no clinical evidence of disease recurrence.

Discussion

MBM of the peritoneum is a rare tumor that occurs predominantly in women of reproductive age (median age, 28 years; range, 18 to 54 years) with only 17% of patients being men [2,3]. One case has been reported in a 9 month old infant [2]. Clinically, there is no specific symptomatology and MBM may even be asymptomatic; the most frequent complaint is chronic pain [3]. MBM, due to compressive effects, presents with abdominal pain and distention and other mass symptoms such as pelvic peritoneum and has a predilection for serosal surfaces of the pelvic viscera. In 1928, Plaut first described the condition as cysts of the pelvis incidentally discovered during surgery for uterine leiomyomas.

The etiology of MBM is controversial but is probably either neoplastic or reactive because of the strong association in women to surgical intervention, inflammatory disease or endometriosis. In general, there is no association with asbestos exposure. These findings along with the predominant benign behavior of MBM support the reactive nature of lesions as opposed to a neoplastic origin [3]. However, recurrence of MBM is as high as 75% and may occur from 1 month to 16 years [4-6]. No risk factors including size, tumor bulk or adequacy of initial resection have been helpful in predicting which tumors will recur [4]. Malignant degeneration is rare but has been reported in 2 of 130 adults [7].

On radiologic examination, MBM appears as intra-abdominal lesions [1,2]. Preoperatively, solitary or multilocular, thin-walled cysts containing watery secretions and multiseptated

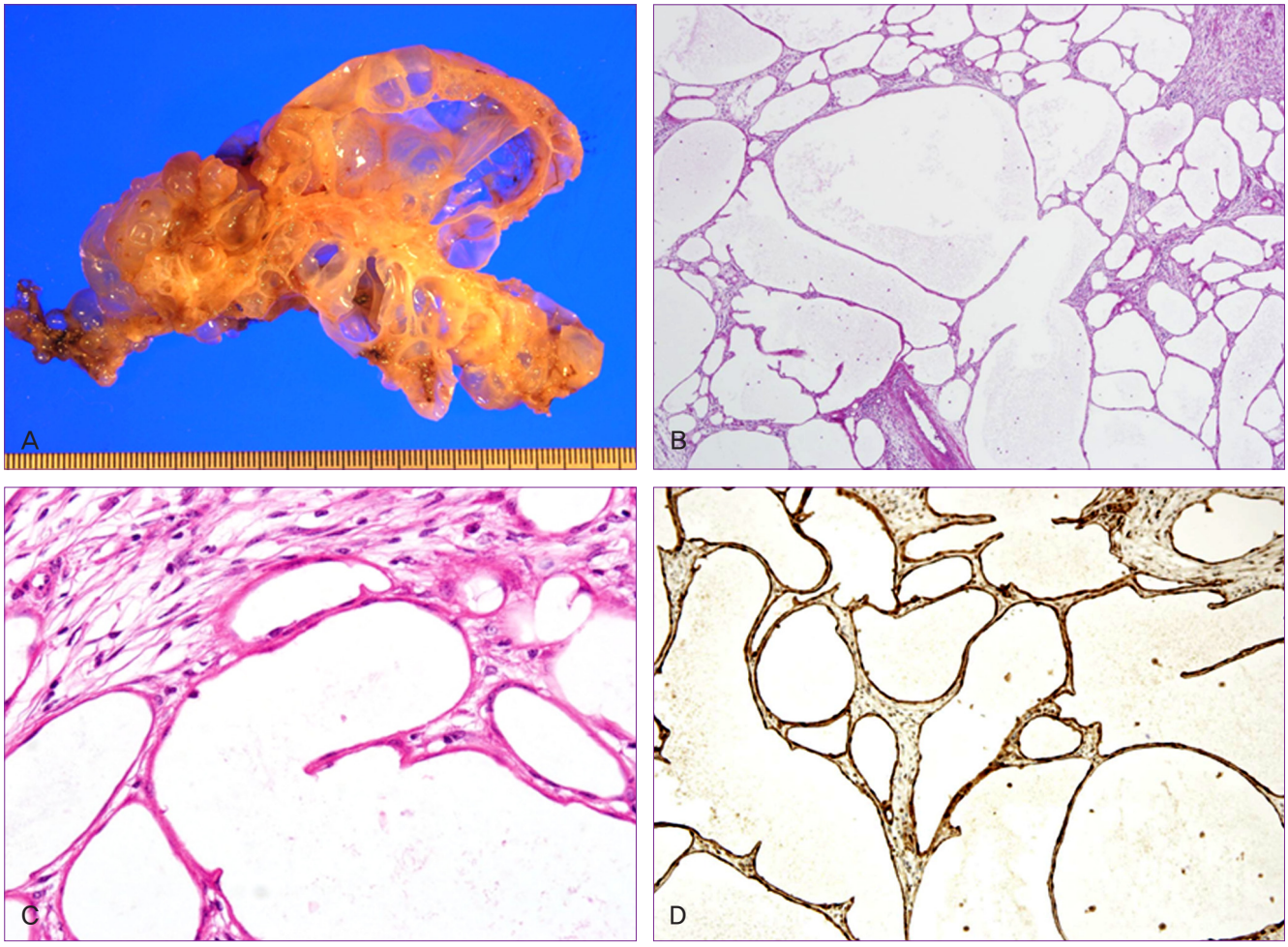

Fig. 2. (A) Multiple transparent cysts. (B) Multiple cysts resembling a lymphangioma (H&E, ×40). (C) The flat shape of the mesothelioma lining the cyst (H&E, ×400). (D) The lining cells are diffuse positive for calretinin (×400).

anechoic cysts, on ultrasonographic examination, point to the presence of MBM [1]. Computed tomography and magnetic resonance imaging confirm ultrasonographic findings but do not permit differentiation from other cystic lesions [1]. The clinical, radiological and morphologic differential diagnosis of MBMs covers the whole range of cystic diagnoses as imaging is not specific. The most reliable method of preoperative diagnosis appears to be cellular analysis of peritoneal washings which has been recently reported to be diagnostic for MBM when abundant mesothelial cells are demonstrated [8]. This analysis should be performed either preoperatively on aspirated peritoneal fluid or on surgically obtained cystic fluid as an adjunct for diagnosis.

The cysts are translucent and arranged in grapelike clusters separated by fibrous tissue. The tumor is multicystic with the cysts ranging in size from several millimeters to as much as 20 cm [1].

Although MBM has not been shown to have malignant potential, operation with complete surgical resection is the recommended treatment. Advantages of resection include confirmation of the diagnosis, ruling out the possibility of malignancy and amelioration of symptoms. Therefore, the treatment of choice, including recurrences, is complete surgical resection. In the present case, laparoscopic approach enabled not only histologic diagnosis but also surgical treatment. Other modalities have been attempted for treating recurrences. Based on the predominance of MBM in women, successful treatment with tamoxifen has been reported in single cases leading to a reduction of the cyst volume and cyst growth [9]. Unfortunately, only about one-fifth of the MBMs are immunoreactive for steroid hormone receptor analysis [3]. One adult patient was treated with intra-ab-

dominal tetracycline through a closed drain following several surgical resections with no recurrence in a 2-year follow-up [10]. Adjuvant chemotherapy and radiation is not warranted because the tumor has a prevailing benign behavior [7]. Rare cases of malignant transformation may be treated with optimal cytoreductive surgery and adjuvant hyperthermic intra-peritoneal administration of cisplatin and doxorubicin [11]. Due to the relatively high frequency of recurrence and possible malignant transition, we recommend a close clinical follow-up of affected patients. Lifelong follow-up is planned with serial physical examinations and imaging using ultrasound for documentation and computed tomography for preoperative localization.

In summary, we wish to alert gynecologists of the diagnostic and therapeutic approach to MBM which can be accomplished by laparoscopy.

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