

Case Report

Benign multicystic peritoneal mesothelioma as a rare disease in an incidental imaging finding in a middle-aged woman: case report and literature review

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ABSTRACT

Benign multicystic peritoneal mesothelioma (BMPM) is a rare, benign cystic neoplasm arising from the peritoneum, most frequently diagnosed in premenopausal women. Fewer than 200 cases have been reported worldwide. Risk factors include endometriosis and pelvic inflammatory disease in women, and prior abdominal surgery in both sexes. Due to its rarity and lack of specific clinical presentation, preoperative diagnosis is challenging, with most cases discovered incidentally. Diagnosis is confirmed through surgical sampling and histopathological analysis. Several treatment strategies have been proposed, with cytoreductive surgery being the standard approach. Long-term follow-up is essential due to the high recurrence rate and the rare possibility of malignant transformation.

Keywords: Benign multicystic peritoneal mesothelioma, Peritoneal neoplasms, Incidental finding, Cytoreductive surgery

INTRODUCTION

Peritoneal mesothelioma represents approximately one-third of all mesotheliomas, the majority of which are pleural.¹ It is conventionally classified into diffuse malignant peritoneal mesothelioma (DMPM) and borderline forms, including benign multicystic peritoneal mesothelioma (BMPM) and well-differentiated papillary peritoneal mesothelioma (WDPPM).¹

BMPM was first described macroscopically by Plaut in 1928 during a procedure for uterine leiomyoma, and histologically defined by Mennemeyer and Smith in 1979.² It accounts for only 3–5% of all peritoneal mesotheliomas. Due to its rarity, available data are limited to case reports and small series.^{3,4}

BMPM predominantly affects women of reproductive age, with a mean age at diagnosis of 42 years and a female-to-male ratio of approximately 4.7:1.⁴ Its pathogenesis remains unclear.⁵ Risk factors include endometriosis, pelvic inflammatory disease and previous abdominal surgery, suggesting chronic peritoneal irritation as a contributing factor.^{4,7} Debate continues over whether BMPM is a reactive inflammatory process or a true neoplasm.^{4,5}

Given the high recurrence rate and potential for malignant transformation, an aggressive treatment approach and lifelong follow-up are recommended upon diagnosis.^{4,5,7,8}

CASE REPORT

A 54-year-old woman with no prior surgical or medical history presented for a general surgery appointment following the incidental discovery of a 38.17×51.53 mm lesion adjacent to the cecum on CT and MRI, initially suspected to be an endometrioma. The case was discussed by a multidisciplinary team of surgical and gynecological specialists and the patient was proposed for an exploratory laparoscopy.

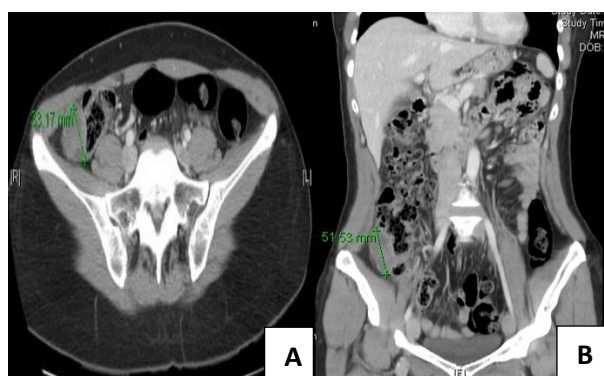


Figure 1: BMPM. The CT image with IV contrast revealed incidental discovery of a 38,17×51,53 mm lesion adjacent to the cecum: (A) axial view CT scan and (B) coronal view CT scan.

The operation was performed and, intraoperatively, a lesion of uncertain origin was found adherent to the cecal wall, along with a mucinous lesion of the ileocecal appendix. Due to the uncertain etiology, a right hemicolectomy was performed to resect both lesions.

Histopathological analysis of the surgical specimen revealed a normal appendix and identified the lesion as a benign multicystic peritoneal mesothelioma. The diagnosis was supported by the presence of an inflammatory polymorphic infiltrate composed of lymphocytes, eosinophils, and binucleated histiocytic cells.

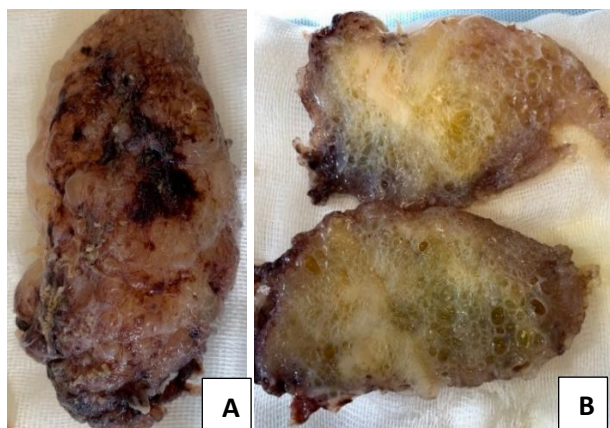


Figure 2 (A and B): Macroscopic image of a benign multicystic peritoneal mesothelioma.

DISCUSSION

BMPM is an extremely rare condition characterized by the proliferation of mesothelial cell-lined cysts in the abdominal cavity.² Most of the available data come from case reports, given the condition's low incidence.^{3,4} Although often asymptomatic, BMPM may present with abdominal pain and distension, tenderness, infertility, or even a palpable mass.^{4,5} Less common symptoms include abnormal uterine bleeding and dyspareunia.⁶ In rare cases, it can lead to bowel obstruction or cachexia.

Preoperative diagnosis is often inconclusive with clinical and imaging data alone.⁴ Ultrasound (US) is typically the first diagnostic tool, revealing multiloculated anechoic cysts.⁴ CT scans help evaluate lesion size and its distribution, typically showing low-density, thin-walled multicystic masses.⁴ However, US and CT are not specific enough to differentiate between BMPM and other cystic lesions.⁴

Magnetic resonance imaging (MRI) is considered the gold standard for the diagnosis.⁴ BMPM appears hypointense on T1-weighted images and hyper- to intermediate-intense on T2-weighted images, with mild contrast enhancement of the cyst walls.⁴ Lymphangioma, primarily seen in children, is an important differential diagnosis.⁴

Most patients remain asymptomatic until the lesion becomes large enough to compress adjacent structures.² Definitive diagnosis requires histological confirmation, which can be achieved by surgery.^{2,5} BMPM is characterized histologically by mesothelial-lined cysts with bland cuboidal cells and clear fluid content.² The surrounding stroma may show an inflammatory infiltration.²

BMPM etiology is still debated.^{4,5} While some believe it is a reactive process linked to inflammation or previous surgeries, others support a neoplastic origin, especially in light of its recurrence rate and rare malignant progression.^{4,5}

The mainstay of treatment is complete surgical excision.^{2,5,8} Hyperthermic intraperitoneal chemotherapy (HIPEC) may improve outcomes, even in asymptomatic cases.^{2,8} Alternative treatments, including hormonal therapy, sclerotherapy, and laser vaporization, have been proposed but lack sufficient evidence.⁶

Recurrence is common and may lead to treatment failure.^{3,5} Due to the risk of recurrence and potential malignancy, long-term follow-up is required for these patients.^{4,5,7}

CONCLUSION

Benign multicystic peritoneal mesothelioma is a rare entity that presents a significant diagnostic challenge due

to its non-specific clinical and radiological features. This case underscores the critical role of complete surgical excision and histopathological examination for definitive diagnosis, particularly when imaging mimics other common appendiceal or gynecological pathologies. By documenting this incidental presentation and its successful management without recurrence over a five-year period, this report enriches the limited existing literature on BMPM, reinforcing the necessity of multidisciplinary approaches and long-term follow-up to mitigate the risk of recurrence and optimize patient outcomes

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