

Case Report

Incidental Finding of Benign Multicystic Peritoneal Mesothelioma: A Case Report

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Abstract

Introduction: Benign multicystic peritoneal mesothelioma represents a rare benign variant of peritoneal mesothelioma, with fewer than 150 cases reported to date. Malignant transformation may occur. We present a patient with an incidental finding of an intra-abdominal mass consistent with benign multicystic peritoneal mesothelioma.

Case presentation: A 51 year-old male presented to the ED with traumatic injuries. During workup, calcified cystic lesions in the pelvis were incidentally noted. Final pathology favored a diagnosis of benign multicystic mesothelioma of the peritoneum. Follow-up imaging obtained three months post-operatively revealed no evidence of recurrent or metastatic disease.

Conclusion: Multicystic peritoneal mesothelioma has been identified as a distinct subtype of peritoneal mesothelioma, with long-term survival achievable through the use of cytoreductive surgery and HIPEC. Although prognosis is relatively favorable, recurrence rates are high, with low potential for malignant transformation. Post-operative surveillance with routine imaging is warranted.

Keywords: Benign Multicystic Peritoneal Mesothelioma; BMPM; Peritoneum

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Consent: Consent was taken from the patient for publication of this case report.

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Introduction

Benign multicystic peritoneal mesothelioma represents a rare and often benign variant of peritoneal mesothelioma, with fewer than 150 cases reported since its first description in the literature in 1979. Malignant transformation may occur. We present a patient with an incidental finding of an intra-abdominal mass consistent with benign multicystic peritoneal mesothelioma.

Case Presentation

A 51 year-old otherwise healthy male presented to the ED with shoulder and leg pain following a bicycle vs. automobile accident. CT scan obtained during routine work-up revealed numerous traumatic injuries (clavicle fracture, scapula fracture, rib fractures, multiple abrasions) as well as incidental calcified cystic lesions in the pelvis (Fig. 1). During his initial hospitalization he recovered uneventfully, and consultation was made for outpatient workup of his intra-abdominal mass.

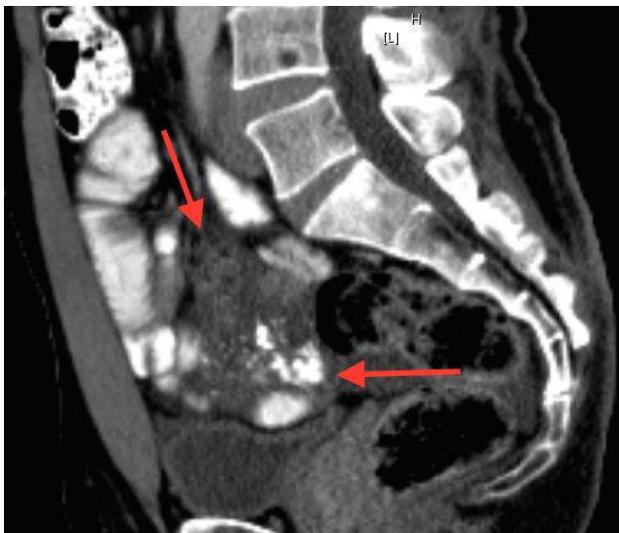


Figure 1 (left): Preoperative sagittal CT image demonstrating a low midline heterogeneously calcified mass

Figure 2 (right): Intraoperative finding of multicystic mass

At the time of his initial evaluation, he endorsed no pertinent abdominal signs or symptoms including pain, anorexia, nausea, or an alteration in his bowel habits. Past medical, surgical, social and family histories were unremarkable and he took no home medications. Of note he had recently undergone routine colonoscopy at an outside institution which was without significant findings. Working differential diagnosis based on imaging included lymphoma vs. low-grade appendiceal malignancy vs. other unspecified pathology. Percutaneous biopsy of the lesion noted only rare atypical cells in a background of mucinous material. Based on these findings and high clinical suspicion for malignancy, the decision was made to pursue operative resection both for cytoreduction and for determination of a definitive diagnosis.

The patient underwent exploratory laparotomy. On opening of the abdomen, he was found to have a 10 x 8 cm multi-lobulated milky white mass localized to the mid-distal jejunum (Fig. 2), with multiple mucinous cystic lesions additionally found throughout the greater omentum and the pelvis. Notably, his appendix appeared normal, though surrounded by mucin. The jejunal mass with adherent

small bowel and urachus was resected en bloc, followed by reconstruction with primary hand-sewn anastomosis. As intraoperative pathologic consultation revealed no evidence of malignancy on frozen section, the decision was made not to utilize hyperthermic intraperitoneal chemotherapy (HIPEC). His cytoreduction additionally entailed greater omentectomy, pelvic peritonectomy, appendectomy and excision of multiple small bowel lesions. Copious hydrogen peroxide irrigation was performed prior to abdominal closure. Post-operatively the patient recovered without incident, and was discharged home on post-operative day six tolerating a regular diet and with full return of bowel function.

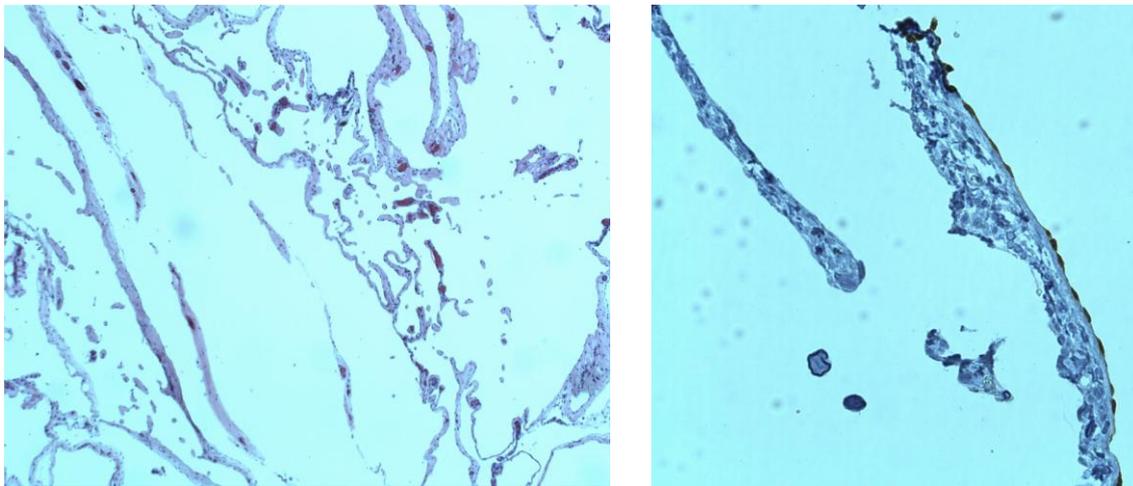


Figure 3 (left): Multiple thin-walled inclusion cysts

Figure 4 (right): Positive calretinin staining of cyst walls consistent with mesothelioma

Final surgical pathology noted a 4 cm fibrocystic mass with multifocal calcification involving the full thickness of the bowel wall. The sampled lymph node was negative and the appendix was benign. The tumor specimen demonstrated multiple inclusion cysts (Fig. 3) lined by a single layer of flat cells which stained positive for calretinin, and negative for CK 5/6, CD31 and CD34 (Fig. 4). Overall there was no evidence of malignancy. Final diagnosis was benign multicystic peritoneal mesothelioma.

Follow-up imaging obtained three months post-operatively revealed no evidence of recurrent or metastatic disease.

Discussion

Benign multicystic peritoneal mesothelioma is a distinct subtype of peritoneal mesothelioma. It is generally considered to most commonly affect young to middle-aged females, though multiple case reports describe occurrences in males [1-3].

Common presenting symptoms include pain, abdominal fullness, and a palpable mass [4-5]. Many, however, are asymptomatic, and are incidental discoveries on imaging or intraoperatively [6]. If preoperative workup is pursued, CT, ultrasound and MRI may help with lesion localization and characterization, but cannot definitively distinguish benign multicystic peritoneal mesotheliomas from other cystic lesions [7-9]. If the mass is amenable, FNA may be performed, but typically show only benign mesothelial cells [10-11].

Theories regarding etiology have been presented, including hormone responsiveness and inflammatory reaction to a previous operation or an intraabdominal process such as endometriosis [1, 5, 12]. It is not thought to be associated with asbestos exposure. Histopathologically, tumors consist of small cysts lined by flattened to cuboidal mesothelial-like cells, with cyst contents consisting of thin watery secretions [4].

Long-term survival is generally achievable through surgical resection, with best results reported with the use of complete cytoreductive surgery and HIPEC [2, 12-15]. Discussion in the literature favors an open surgical approach, though laparoscopic approaches have been described [16].

Conclusion

The natural history of benign multicystic peritoneal mesothelioma is poorly understood [17-18]. Although prognosis is relatively favorable, recurrence has been reported with rates as high as 27-75% [5, 19, 20]. Potential for malignant transformation has also been described [2, 21]. Although metastasis is typically not noted, many authors question whether such tumors are truly benign [21, 22]. Post-operative surveillance with routine imaging is therefore warranted, though there exists no consensus regarding frequency, duration or modality of imaging.

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