



BENIGN MULTICYSTIC PERITONEAL MESOTHELIOMA IN MALE: A CASE REPORT

Oncology

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ABSTRACT

Benign Multicystic peritoneal mesothelioma (BMPM) is a rare benign neoplasm, mostly found in premenopausal females. Aetiopathogenesis is unclear, but it may be linked to excessive inflammatory process. We are reporting a rare case of Benign Multicystic Peritoneal Mesothelioma in male, who presented to us with complain of pain abdomen and a soft, cystic mass on abdominal examination. He had history of appendectomy 7 years ago. Patient underwent cytoreductive surgery including total omentectomy with right hemicolectomy. Due to rarity of disease, a definitive treatment protocol and prognosis has not been established so far. Currently a complete cytoreduction surgery (CRS) with hyperthermic intraperitoneal chemotherapy (HIPEC) remains the most widely accepted treatment to reduce the recurrence rates.

KEYWORDS

Benign Multicystic peritoneal mesothelioma, Mesothelioma, CRS, HIPEC

INTRODUCTION

Benign Multicystic Peritoneal Mesothelioma (BMPM) is a rare neoplasm with approximately 200 cases reported in literature till date. [Noiret B, et al 2019 ; Kelarji AB et al 2021] It was first identified in 1928 by Plaut, and its histological description was given by Menemeyer and Smith in 1979. [Zahid A et al, 2021] It is mostly found in premenopausal females and rarely reported in males.

It is characterized by multilocular cystic spaces with fibrous walls lined by mesothelial cells, involving pelvis and peritoneal cavity. [Sawh RN, et al, 2003] Seven histological types have been identified thus far: low-grade mesothelioma, well differentiated papillary, multicystic, epithelioid, sarcomatoid, biphasic and deciduioid [Chand MT, et al, 2020].

Various theories suggest association of BMPM with chronic peritoneal inflammation, oestrogen, endometriosis, pelvic inflammatory disease, previous pelvic surgery or abdominal surgery and possibly neoplastic origin, but aetiopathogenesis is still not well elucidated. [Noiret B, et al, 2019 ; Sugarbaker PH, et al 2003]

Due to rarity of disease, a definitive treatment protocol and prognosis has not been established so far. From the available data, it can be concluded that prognosis is good after timely diagnosis and surgical intervention.

CASE REPORT

A 21 year male presented to us in July 2023, with complain of abdominal pain for 4 months with associated swelling in abdomen. He has previous history of appendectomy 7 years ago. Now on examination there was vague intrabdominal mass, which was cystic in consistency. Sonography was suggestive of diffuse complex cystic lesion / septated collection filling the peritoneal space extending from subhepatic space to pelvis. CECT abdomen reported diffuse lobulated hypodense soft tissue filling peritoneal cavity with prominent vessel in the enhancing septations. CT chest was unremarkable, routine hematology was within normal parameters and CEA & CA 19.9 were within normal range.



Figure 1- Intra-abdominal mass containing multiple cyst on Laprotomy

On laparotomy, there were multiple (>1000) cysts with serous and mucin containing fluid involving omentum, peritoneum mesentery of caecum and pelvic peritoneum along with multiple subcentimetric mesenteric lymph nodes and ascites. [Figure-1] For which total omentectomy and right hemi colectomy was done. Post operative period was uneventful.

Histopathological analysis revealed multicystic benign mesothelioma in the omentum, as well as in mesentery of caecum and ascending colon. [Figure-2 &3] Ascitic fluid cytology reported acellular mucin pool without malignant cell with markedly raised LDH. Immunohistochemistry (IHC) was positive for Calretinin and Ki67 was less than 1%. Thus, the diagnosis of Benign Multicystic Peritoneal Mesothelioma (BMPM) was confirmed. Patient is on regular follow up with no evidence of recurrence till date.

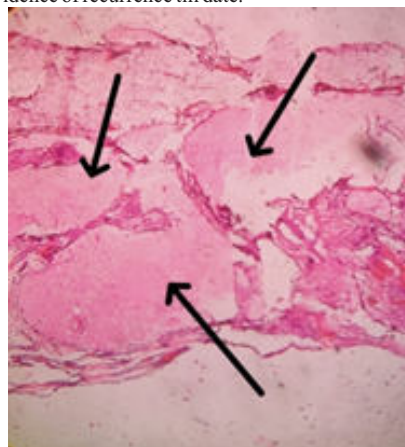


Figure 2 - Arrows Showing Multiple Cysts

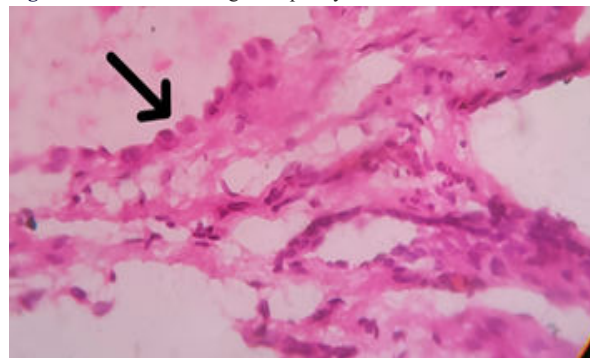


Figure 3 - Cyst wall lined uniform layer of benign mesothelial cells, no cellular atypia

DISCUSSION

Benign Multicystic Peritoneal Mesothelioma (BMPM) is a rare peritoneal neoplasm and pathogenesis is not clear. Some believe it to be linked to chronic inflammation in peritoneum leading to reactive hyperplastic and dysplastic changes of mesothelial cells. Others suggest primitive neoplastic origin from peritoneal serosa without association of inflammation. [Zahid A, et al, 2021] Mostly it is reported in female of reproductive age group.

Clinically disease is usually vague, mostly asymptomatic in early stage and diagnosed incidentally on abdominal exploration or Laparoscopy. It may become symptomatic once it involves other organs like bowel, bladder and kidney. Symptoms may include chronic or intermittent abdominal pain, abdominal fullness, change in bowel and bladder habit, abdominal distension, intestinal obstruction and urinary retention. Physical examination may reveal abdominal distension, tenderness and a vague abdominal mass may or may not be extending to the pelvis. It may simulate other intrabdominal pathologies like appendicitis, intestinal obstruction, irritable bowel disease etc.. Sonography finding of multiple cystic masses should raise a suspicion of benign multicystic peritoneal mesothelioma.

Differential Diagnosis of BMPM includes Cystic Lymphangioma, Endometriosis, Adenomatoid tumours, Serous tumours of peritoneum, Malignant Mesothelioma and Ovarian Clear Cell Carcinoma. [Sugarbaker PH, et al, 2003]

BMPM is considered a borderline malignancy, which is prone to multiple recurrences, and few reports suggest development of squamous metaplasia and malignant transformation. [Zahid A, et al, 2021] During laparotomy for other causes if multicystic lesion is found, a possibility of BMPM should be considered during histopathological assessment.

Mainstay of the treatment is a complete and through cytoreduction, but it has a recurrence rate of 40-50%. [Zhang CH, et al, 2017] To address this issue multiple treatment modality like adjuvant systemic / intra-abdominal chemotherapy, abdominal wall radiotherapy, hormone (anti estrogen/ GnRH agonist) therapy, sclerotherapy and laser vaporization have been attempted. Currently a complete cytoreduction (CRS) with hyperthermic intraperitoneal chemotherapy (HIPEC) remains the most widely accepted treatment. This modality reduces the rate of recurrence to 20% and results in 10 year survival rate of 100%. [Zahid A, et al, 2021]

CONCLUSION

Benign Multicystic Peritoneal Mesothelioma (BMPM), mostly affects female of reproductive age group. Our case is a Benign Multicystic Peritoneal Mesothelioma (BMPM) in a young male, which is a rare disease. High index of suspicion must be there when any patient present with vague abdominal pain and mass along with multiple cystic lesion on sonography. Although histologically benign, it is prone to multiple recurrences. Early surgical intervention with hyperthermic intraperitoneal chemotherapy (HIPEC) has been reported to result in a better prognosis.

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