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A Rare Case of Benign Cystic Peritoneal Mesothelioma.

KS Ravishankar, R. Kannan, Srivatsan Gurumoorthy, Gokul D Yatheendranathan*, and Rajesh Gandhi.

Department of General Surgery, Sree Balaji Medical College & Hospital, Bharath University, Chrompet, Chennai – 600044, Tamil Nadu, India.

ABSTRACT

Benign cystic peritoneal mesothelioma is a rare clinical entity with variety of presentations. Association with various intraabdominal conditions makes the diagnosis difficult. Herewith we report a case of benign cystic peritoneal mesothelioma presenting as an acute abdomen.

Keywords: Benign cystic peritoneal mesothelioma, mesothelial cells.

Corresponding author

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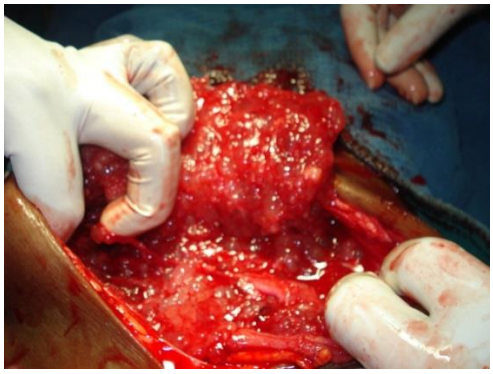
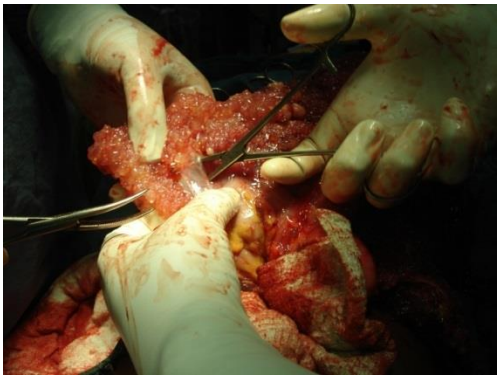
Case report

A 13 year old girl presented to the emergency department with history of sudden onset of lower abdominal pain for 3 days. There was history of 1 episode of vomiting which was not blood or bile stained. She did not have fever, dysuria, or bowel disturbances. Examination revealed tachycardia, moderate pallor, lower abdominal guarding and sluggish bowel sounds. Per vaginal & per rectal examinations were normal.

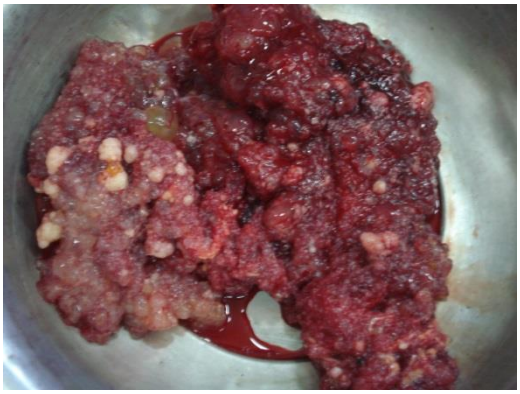
USG abdomen revealed 16 x 13.4 x 6.7 cm sized echogenic irregular ill-defined mass with cystic spaces, septations & calcifications, noted arising from the pelvis and extending into the supraumbilical region in the midline. Uterus was normal and ovaries could not be delineated clearly. An emergency plain abdominal CT confirmed the above findings.

Patient was adequately hydrated, analgesics and antibiotics were administered. Since the patient had persistent tachycardia and lower abdominal guarding, we decided to proceed with laparotomy.





Multiple cystic clusters arising from the pelvis and involving the entire peritoneal cavity



Excised specimen

Laparotomy showed a large mass of size 15 x 13 cm arising from the pelvis extending up to the umbilical region. It was predominantly cystic aggregate, the cysts being composed of clear, watery fluid. The cystic mass, resembling bunch of grapes was seen to involve the anterior abdominal wall, pelvis, ovaries, uterus, mesentry of small bowel, transverse mesocolon and appendix. However there was no luminal narrowing of small or large bowel. Stomach, duodenum, pancreas, spleen and liver were normal. No para-aortic or mesenteric were visualised.

A debulking surgery was done removing the bulk of the tumour from the peritoneum. Post-operative period was uneventful and the patient was discharged on 10th post op day. Patient made an excellent recovery after surgery.

The histopathological report of the resected specimen was “Benign Cystic Peritoneal Mesothelioma”. Further search into the literature revealed that it was an extremely rare entity.

RESULTS AND DISCUSSION

Benign Cystic Peritoneal Mesothelioma is an extremely rare entity. Only about 130 cases have been reported in literature worldwide.

In 1889 Henke [1] reported a case of "multiple cystic lymphangioma like tumour". Over the next hundred years various similar growths were described and it was only in 1980 that the now commonly accepted term, "benign cystic mesothelioma", was used [2]. The mesothelial origin of such growths had been demonstrated by Mennmeyer and Smith in 1979 [3].

It is also known as benign multicystic peritoneal mesothelioma (BMPM) or Multilocular peritoneal cysts.

BCPM is a localized tumor arising from the epithelial and mesenchymal elements of the mesothelial cells, and does not metastasize. The tumor can attach to serosal surfaces of the intestine and omentum or in the retroperitoneal space, spleen and liver if located in the peritoneal cavity [7]. The aetiology remains obscure. In particular there is no association with asbestos exposure, previous abdominal surgery, or abdominal trauma [4, 6].

It shows predominant female preponderance, especially those in reproductive age group. Any cause of peritoneal irritation like pelvic inflammatory disease, endometriosis, ruptured ectopic pregnancy, SLE may lead to this condition. The most common presenting complaints are pelvic and low abdominal pain, but the lesions are sometimes incidental findings at laparotomy. Rarely, weight loss and a huge abdominal mass can be the presenting features.

Ultrasonography can give useful information but computed tomography scanning is the investigation of choice. With this imaging modality the differential diagnoses include peritoneal lymphangioma, pseudomyxoma peritonei, and cystadenoma or cystadenocarcinoma of the ovary.

Aspiration cytology can be useful in making a preoperative diagnosis [1]. Typically, the aspirate from a BCPM contains mesothelial cells showing focal presence of a brush border epithelium. Immunohistochemistry is also helpful in problematic cases [7,8].

Surgery is the definitive treatment, a debulking procedure is usually sufficient. Grossly, the cysts filled with mucinous or gelatinous fluid range from several millimeters to more than 20 cm in diameter. Characteristically, the tumor is composed of a multiple mesothelial-lined cystic structure, with fragile fibrovascular stroma holding the formation together. It is not responsive to chemo or radiotherapy. Prognosis is excellent, though there are chances of recurrence. Periodic follow up of at least once in 3 months for first 2 years, 6 months in next 2 years and yearly thereafter is needed. The reported recurrence rate is slightly higher in women (40-50%) than in men (33%) [5].

Cystic lymphangioma (cystic hygroma), cystic adenomatoid tumor, cystic forms of endosalpingiosis, endometriosis, Müllerian cysts involving the retroperitoneum, and cystic mesonephric duct remnants are

included in the differential diagnosis among benign lesions. Malignant lesions include malignant mesothelioma and serous tumors of the peritoneum.

Our patient reported twice for follow up in the last 6 months and there has been no clinical or radiological evidence of recurrence and patient is able to carry out her activities without any difficulties.

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