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Case Report

Benign cystic peritoneal mesothelioma

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ABSTRACT

A well-defined but rare entity of Benign Cystic Peritoneal Mesothelioma (BCPM) is reported. The aetiology of this neoplasm remains obscure. The presenting features make a precise preoperative diagnosis difficult but information provided by computed tomography and cytology may help. A firm diagnosis can only come from an electronic microscopy or immunohistological examination of the tumour. Diagnostic accuracy and diligent follow up are essential because, although the tumour is considered benign, it does tend towards local recurrence.

Keywords: BCPM, Cytokeratin stain, Recurrence, Resection, Marsipulation

INTRODUCTION

Benign Cystic Peritoneal Mesothelioma (BCPM) is a neoplasm composed of multiple fluid filled cysts. In terms of malignant potential it lies in between two other neoplasm of mesothelial origin: adenomatoid tumour and malignant peritoneal mesothelioma. The more common peritoneal adenomatoid tumour is usually an incidental finding at laparotomy and rarely causes symptoms. In contrast, the malignant mesothelioma, that is known to develop after exposure to asbestos, frequently metastasis and is fatal. BCPM does not metastasise and therefore is not truly malignant but frequently recurs locally. BCPM is more common in women of reproductive age, there have been only 20 cases reported in men.

CASE REPORT

A 49 year old man presented with intermittent generalised abdominal pain of three years duration. Three months before admission he had noticed a swelling on the right side of his abdomen which had grown steadily in size. There were no other symptoms.

On examination, there was a large lobulated mass with well-defined margins occupying the entire right side of abdomen extending into the hypogastrium. A small mass with similar features was palpable in the left iliac fosse. There was no hepatosplenomegaly or ascites.

Serum biochemistry was normal.

Ultrasonography showed a large intraperitoneal multicystic mass not obviously arising from any solid organ. Ultrasonographically guided needle aspiration yielded pale yellow fluid which showed mesothelial cells on cytology.

At laparotomy the multicystic mass was found to occupy the entire abdomen and was adherent to small bowel, large bowel, liver and bladder (Figure 1 and 2). The tumour was resected except where inseparable from viscera.

The patient made an uneventful post-operative recovery and no further treatment is planned at the time of writing. Three months after surgery patient remained asymptomatic.

Grossly, the specimen was a gelatinous mass. Its cut surface showed multiple cystic spaces.

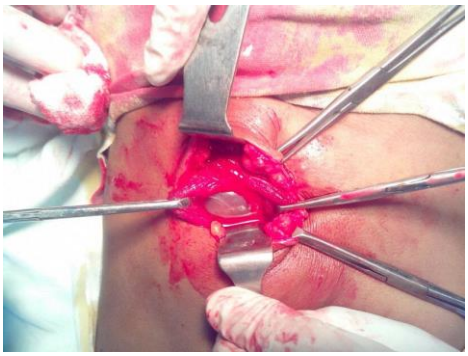

Figure 1: The multicystic mass at laparotomy.

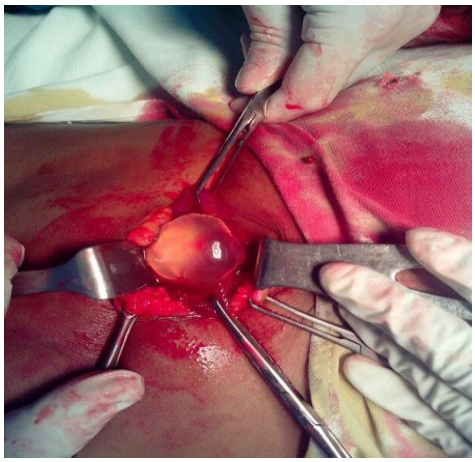

Figure 2: Occupation of the entire abdomen with the multicystic mass at laparotomy.

Microscopically, the tissue showed cystic spaces of varying sizes separated by fibrous septa. The spaces were lined by single layer of flattened or cuboidal cells. On immunohistochemical staining, there were cells that stained positive for cytokeratin (CAM 52) and negative for Factor VIII related antigen, thereby confirming there mesothelial origin (Figure 3 and 4). The histological diagnosis was benign cystic peritoneal mesothelioma.

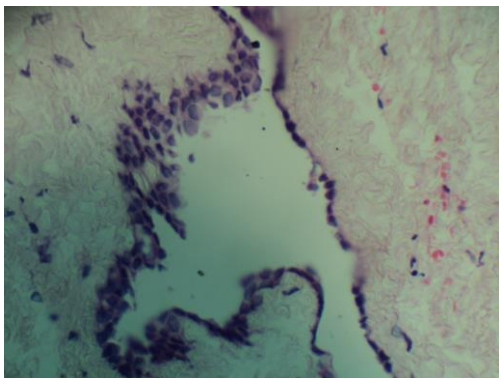

Figure 3: Immunohistochemical staining - I.

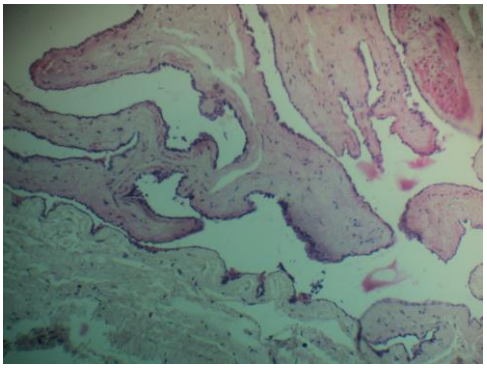

Figure 4: Immunohistochemical staining - II.

DISCUSSION

The clinical and pathological findings described in relation to our case represent typical features of BCPM.

In 1889, Henke[3] 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. The mesothelial origin of such growth had been demonstrated by Mennmeyer and Smith in 1979.[5] The aetiology remains obscure. In particular there is no association with asbestos exposure, previous abdominal surgery or trauma.

Abdominal pain, tenderness and distension, usually in association with a pelvic or abdominal mass are the most common presenting features. Ultrasonography can give useful information but computed tomography is the investigation of choice. With this imaging modality the differential diagnosis include peritoneal lymphangioma, pseudomyxoma peritonei and cystadenoma or cystadenoma of ovary in females.

Aspiration cytology can be useful in making a preoperative diagnosis. Typically, the aspirate from a BCPM contains mesothelioma cells showing focal presence of brush border. Conditions such as cirrhosis of liver, viral infections and connective tissue disorders with mesothelial hyperplasia can also yield an aspirate with mesothelial cells. But in these conditions the cells tend to form clusters and aggregates as opposed to large flat sheets seen in BCPM.

At surgery the tumour typically consists of multiple, translucent cyst forming a confluent mass. The tumour has been reported to include various parts of the serosa of the bowel and sometimes spreads to the liver, spleen and the pancreas. Rarely the tumour may present as free floating pelvic cysts.[6] Histopathologically, BCPM is composed of multiple mesothelial lined cystic spaces separated by a delicate fibromuscular stroma. Mild to moderate inflammatory changes are occasionally seen.

The mesothelial origin is confirmed by electron microscopy and immunohistochemistry. The former show the characteristics of mesothelioma cells-slender microvillus on the luminal surface of the cells, desmosomes, intracytoplasmic intermediate filaments, endoplasmic reticulum and mitochondria. The later shows strong staining for cytokeratin in the cystic linings and for vimentin in subepithelial cells. Absence of both these features is otherwise similar tissue suggests the diagnosis of lymphangioma.[7]

Special stains (Masson trichrome) can be used to demonstrate the presence of muscle fibres in the cystic lymphangioma. These muscle fibres are absent from the walls of BCPM. Adenomatoid mesothelial tumours and the malignant mesothelioma can easily be differentiated from BCPM by their gross and histological characteristics. Accurate diagnosis of BCPM is important because, though benign, these tumours tend to recur locally.

BCPM is treated by surgical resection. When complete removal is impossible due to adherence, marsupialisation of the remaining cyst is recommended. Radical resection is not advocated if it puts vital structures to risk. Intraperitoneal tetracycline has been used to sclerose residual tumour after surgical resection,[8] but there is no evidence that this is beneficial. Chemotherapy and radiotherapy are of no value. Considering the potential of this tumour to recur, close follow up is essential. Substantial recurrence usually warrants further resection.

The prognosis of BCPM is excellent. The reported recurrence rate is slightly higher in women (40-50%) than in men (33%).[2] Surprisingly, there has been one death reported from BCPM, the patient dying as a result of local tumour effects 12 years after refusing surgery.[7]

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