Benign cystic mesothelioma of omentum: Mimicking abdominal lymphangioma

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ABSTRACT

Benign multicystic peritoneal mesothelioma (BMPM) or benign cystic mesothelioma is a very rare disease characterized by proliferative lesions of the peritoneal mesothelial cells mainly in the pelvic or abdominal cavity. It occurs predominantly in young to middle-aged women and the majority of cases were associated with a history of abdominal or pelvic operation, a history of endometriosis, and pelvic inflammatory disease. BMPM involving only the omentum is not mentioned in the literature; here, we present a unique case of BMPM which was diagnosed as cystic lymphangioma with differential diagnosis of pseudomyxoma peritonei in a 40-year-old patient presenting as recurrent abdominal pain.

Key words: *Ascites, Benign cystic mesothelioma, Lymphangioma, Peritoneal carcinomatosis*

Benign multicystic peritoneal mesothelioma (BMPM) was first described in 1979 by Mennemeyer and Smith [1]. It is a rare peritoneal tumor with a strong predilection for the peritoneum of pelvic organs [2]. This tumor is misleading as there are several differential diagnoses such as cystic lymphangioma, pseudomyxoma peritonei, and cystic teratoma which are the most common. It is common in women in their reproductive age group. At present, approximately <200 cases have been reported all over the world. The pathogenesis of BMPM is still controversial regarding its neoplastic and reactive nature. We would like to report this unusual case of BMPM involving the omentum only.

CASE REPORT

A 40-year-old man was admitted to our hospital with a history of repeated upper abdominal pain for 6 months. The abdominal pain was intermittent, dull aching in nature, confined to the epigastric, and the left hypochondriac region. The pain was aggravated with food, relieved by analgesics, and not associated with fever or vomiting. There was no history of altered bowel habits, hematemesis, or melena. There was no similar complaint in the past. He had a previous history of pulmonary tuberculosis 3 years back and took a complete course of antitubercular medication. He is a known alcoholic, taking country liquor 90–180 ml/day. Sleep pattern and appetite were normal and there was no significant weight loss.

At the time of admission, his health status was normal: Blood pressure 139/86 mmHg right arm supine position, pulse rate 73 beats/min, respiratory rate 20/min, and temperature 36.5°C. He had no apparent trace of the disease and was conscious. The breathing sound and the heart sounds during the chest examination were found to be normal. There was a little abdominal distension with a diffuse non-tender lump felt on the epigastric and periumbilical region. The lump was intra-abdominal, firm in consistency and moves with respiration. There was no hepatosplenomegaly and ascites. Hernia orifices were free and bowel sound appears good.

The peripheral blood examination showed normal findings, i.e., hemoglobin – 14.2 gm/dl. leukocytes – 7820/mm^3, and platelets – 220,000/mm^3 with normal renal and liver parameters. Ultrasonography (USG) of the abdomen showed a large, ill-defined abdominopelvic heterogeneous cystic lesion measuring approximately 8.7×11.1×7.1 cm, extending from the left hypochondrium to the pelvis with internal septations. Contrast-enhanced computed tomography (CECT) abdomen and pelvis showed a multiloculated cystic lesion with thin enhancing septa measuring approximately 18×9.5×8.1 cm noted involving the left abdominopelvic region extending from the epigastric to the pelvic region, few specks of calcification noted. On the basis of USG and CECT findings, provisional diagnoses of lymphangioma and pseudocyst of the pancreas were kept (Fig. 1).

We planned for diagnostic laparoscopy for further evaluation. Diagnostic laparoscopy revealed multicystic lesions with clear fluid seen arising from the greater omentum (Fig. 2). On opening lesser sac, there was no pseudocyst component and spleen was congested. Hence, we converted our procedure to midline laparotomy. Laparoscopic findings confirmed and infracolic omentectomy done. The specimen was sent for a histopathological examination.

The patient had an uneventful post-operative period and was discharged in 7 days. Histopathology on gross examination

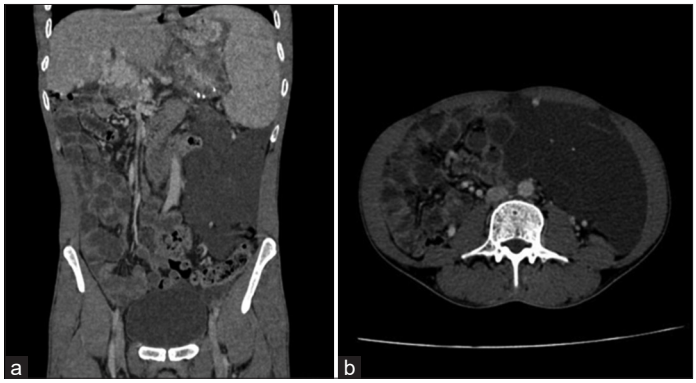

Figure 1: Computed tomography abdomen mass lesion showing (a) left hypochondriac region to pelvis; (b) multiple septation

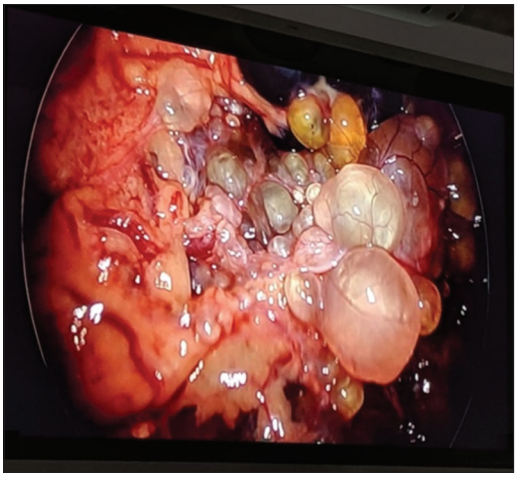

Figure 2: Laparoscopic view omental mass

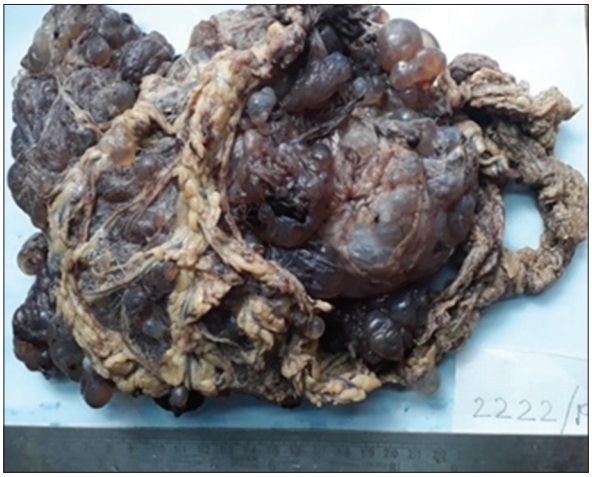

Figure 3: Gross examination showing a single jar containing a mass of omentum with multiple cysts measuring 32.0×12.0×6.0 cm

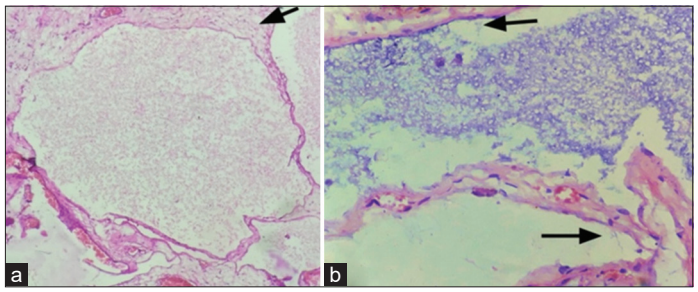

Figure 4: (a and b) Histopathological examination showing cystic spaces separated by fibro-fatty tissue and congested blood vessels; ×1000

revealed omentum with multiple cystic mass of 32×12×6 cm (Fig. 3). Microscopy showed multiple cysts, lined by flattened epithelium, and filled with proteinaceous material. These cysts were closely approximated and of varying sizes. Occasional areas of native fibro-fatty tissue were seen in between the cysts. No cytological atypia or proliferation of the lining epithelium was seen (Fig. 4). On follow-up after 1 and 2 months, the patient was well asymptomatic. USG abdomen done showed mild hepatomegaly.

DISCUSSION

BMPM was first described in 1979 by Mennemeyer and Smith [1]. Since then, approximately <200 cases have been reported. It has a strong predilection for the surfaces of the intestine, pelvic viscera, omentum, retroperitoneal space, spleen, and liver [2]. Our case also showed the origin of the tumor from the greater omentum. The BMPM affects women in 80% of cases, with an average age of 34 years [3]. BMPM is a localized tumor that has a high local recurrence rate [4]. The recurrence rate is higher in women (40–50%) than in men (33%) [5].

The etiology remains unclear; some authors have proposed a neoplastic origin based on slow but progressive growth of the lesions, a tendency to recur after surgical resection. Malignant transformation is very rare [3]. In the literature, only two cases have been reported of malignant transformation.

Ultrasound and computed tomography do not differentiate benign cystic mesothelioma from other cystic lesions. Magnetic resonance imaging (MRI) may confirm the peritoneal origin, differentiate cystic content, or may detect other lesions. In BMPM, typical MRI findings are low signal on T1-WI and high signal on T2-WI with peripheral enhancement following gadolinium administration [6]. Diagnostic laparoscopy is the most accurate method since it allows visualization and local biopsy of the suspected tissue specimens [7].

There are no clear guidelines on the management protocol for BMPM. In the present case, diagnostic laparoscopy was done as there was no clear diagnosis on imaging. Surgery with complete nucleation of the cyst to prevent recurrence and possible malignant transformation remains the mainstay of treatment. However, some authors advocate aggressive surgery followed by heated intraperitoneal chemotherapy (HIPEC) using cisplatin or doxorubicin [8]. Recurrences occur more frequently in women than men as they are treated by hormonal therapy with anti-estrogen [9], gonadotropin-releasing analogs [8], sclerotherapy with tetracycline, and HIPEC. These methods may be efficacious in the treatment of more aggressive disease and it may reduce the need for repeated laparotomies.

CONCLUSION

BMPM is a rare tumor with a high local recurrence rate and requires a high index of suspicion for diagnosis, so we recommend diagnostic laparoscopy whenever the case suspects BMPM, although final diagnosis should be made by histopathology. Long-term follow-up of these patients is required since there is a high recurrence rate and further resection or additional therapy may be required.

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