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Case Report

Benign Multicystic Peritoneal Mesothelioma: A case report

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ABSTRACT

Benign multicystic peritoneal mesothelioma is a well defined but rare neoplasm with a non specific appearance on imaging. The aetiology of this neoplasm remains obscure. The presenting features make a precise preoperative diagnosis difficult. CT and MRI are useful in suggesting the preoperative diagnosis but confirmation of diagnosis requires pathological analysis. We report the case of a patient with benign multicystic peritoneal mesothelioma and describe its appearance on magnetic resonance imaging, in correlation with gross, clinical and pathological findings.

KEYWORDS: Benign multicystic peritoneal mesothelioma (BPM), MR appearance, Pathological findings.

INTRODUCTION

Benign multicystic peritoneal mesothelioma (BPM) is a neoplasm composed of multiple small fluid-filled cysts arising from peritoneal mesothelium. It is often diffuse and shows a marked predilection for the surfaces of the pelvic viscera. [1,2] This disease is a rare medical entity and approximately 130 cases have been described in the literature till date and there are challenges in determining its origin, pathogenesis, diagnosis and therapy.

CASE REPORT

A 38 year old woman was admitted to our surgical department with the complaints of intermittent pain in her lower abdomen. The patient had undergone hysterectomy eight years back. There was no other significant medical or family history. There was no discretely palpable mass on physical examination. Laboratory data were within normal ranges.

Abdominal ultrasonography demonstrated a multiloculated mass in the pelvis, dominantly in the right adnexal area measuring approximately 77 x 68 x 92 mm, not obviously arising from any solid organ. MR study revealed a multiloculated cystic mass lesion in the pelvis measuring 11.4 cm (transverse) x 10.1 cm (anteroposterior) x 8.0 cm (craniocaudal) in size. The lesion showed multiple loculi of varying signal intensity demonstrating predominantly hypointense signal on T1-weighted images and hyperintense signal on T2-weighted images. (Fig.1) The lesion appeared intraperitoneal and located superiorly in the pelvis and posterior to the dome of urinary bladder. The adnexa were not separately visualised and were possibly engulfed within the lesion. The diagnostic possibilities suggested were mucinous cystadenoma of ovary / lympho venous malformation / peritoneal inclusion cyst / pelvic hydatidosis involving the ovaries and fallopian tubes. No significant pelvic lymphadenopathy was seen.

At laparotomy a multicystic mass was found to occupy the entire pelvic cavity engulfing the adnexae and extending up to the vaginal cuff. The mass also extended to the abdominal cavity and was adherent to both large and small gut. On gross examination the specimen showed irregular multilocular gelatinous cystic mass consisting of numerous clear fluid filled cysts ranging in size from 0.3 cm to 4 cm in diameter with a wall thickness of 0.1 cm.(Fig.2) No necrosis, haemorrhage or solid areas were identified. Microscopy revealed a multicystic mesothelioma characterised by multilocular cysts lined by flat to cuboidal epithelium with no malignant features. (Fig.3) The septa consisted of loose connective tissue with scattered lymphocytic infiltrate and vascular congestion. (Fig.4) The mesothelial nature of the lining was confirmed by positive immunohistochemical staining for cytokeratin and calretinin. CD34 was negative in these cells.

Figure: 1 MR image demonstrates a multicystic mass in the pelvic region

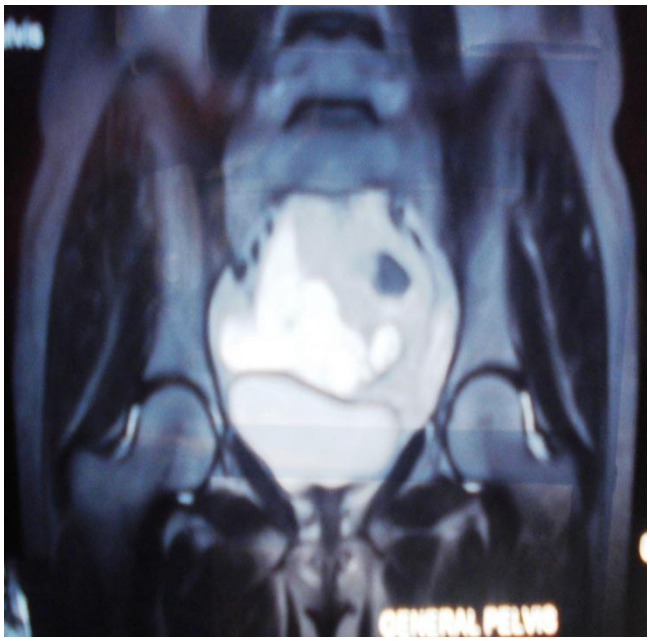


Figure: 2 Photograph of the resected specimen showing multiple cysts with clear serous contents

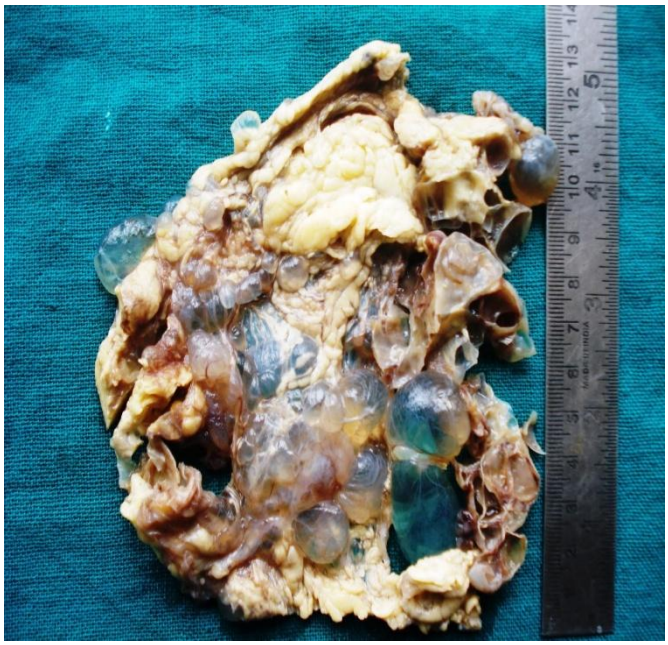


Figure: 3 Photomicrograph showing multicystic nature of the tumour. (Hematoxylin & Eosin stain, 100x)

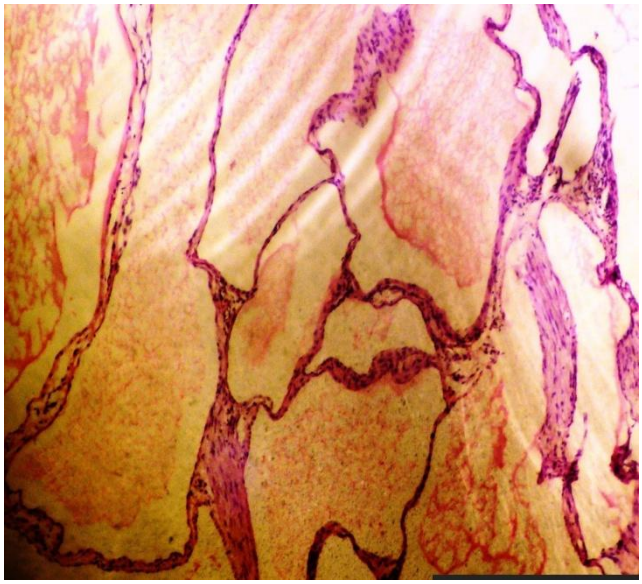
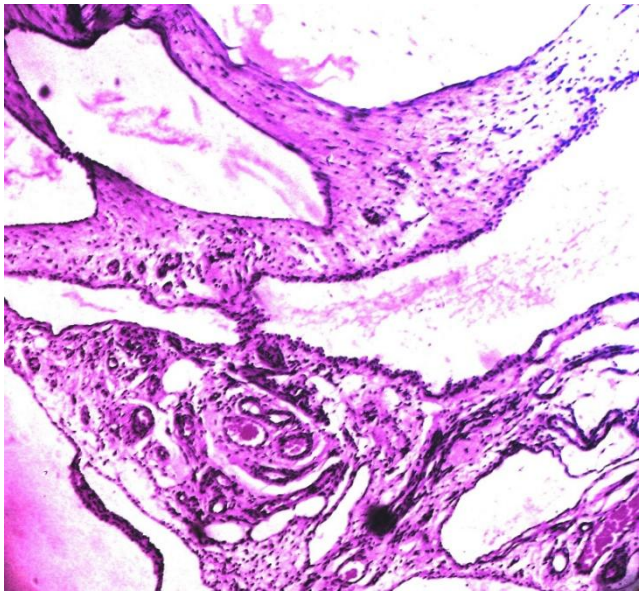


Figure: 4 Photomicrograph showing variable sized cysts lined by flattened mesothelial cells. (Hematoxylin & Eosin stain, 400x)



DISCUSSION

Mesotheliomas are mesenchymal neoplasms originating from the serous lining of the pleural, pericardial or peritoneal space. Multicystic peritoneal mesothelioma involves the peritoneum or extra-peritoneal space, omentum, pelvic or abdominal viscera. It most commonly arises from the pelvic surfaces of the peritoneum and has benign or indolent biologic behavior. [3-5] Multicystic mesothelioma of the peritoneum was first described in 1979 by Mennemeyer and Smith [6] and since then approximately 130 cases have been described in the literature. It is an intermediate-grade tumor, between the benign adenomatoid tumour of the peritoneum and the more common malignant asbestos-related peritoneal mesothelioma. [4]

BPM commonly occurs in young to middle-aged women (mean age, 37 years). The presenting symptoms are chronic or intermittent lower abdominal or pelvic pain, tenderness, or distension with an abdominal or pelvic mass and, less frequently this neoplasm may manifest as an incidental finding. [1,2] Women with this lesion often have a history of

prior pelvic surgery, endometriosis or pelvic inflammatory disease, favouring a reactive nature of this process. [7-9]

Radiologic findings in multicystic mesothelioma include multilocular thin-walled cysts containing watery secretions. US demonstrates multiseptated anechoic cysts. CT demonstrates a well-defined, low-attenuation mass with non calcified septa. Historically, CT has been the most commonly used preoperative modality however, MR imaging provides additional coronal and sagittal planes, which assist in the assessment of both the relationship with bowel and possible visceral organ involvement. [5]

The differential diagnosis includes lymphangioma, other mesenteric-omental cysts, ovarian cystadenoma and cystadenocarcinoma, cystic teratoma, pseudomyxoma peritonei, cystic smooth muscle tumours, visceral cysts, cystic mucinous neoplasms of the pancreas, non pancreatic pseudocysts, and endometriosis. [5] Several of these entities have radiologic features that help differentiate them from multicystic mesothelioma. Lymphangiomas can be radiologically identical to multicystic mesotheliomas; however, they often occur in younger patients and can be identified if they contain predominantly chylous fluid. [10,11]

Mesenteric cysts are generally unilocular and contain serous secretions, with no discernible wall or internal septa. [11] Teratomas contain fat and calcification, which are not present in multicystic mesotheliomas. Non pancreatic pseudocysts may be either unilocular or multilocular. They can be differentiated from multicystic mesothelioma on the basis of their thick wall and varying debris contents (serous, hemorrhagic, purulent). Malignant neoplasms are suggested by ancillary signs such as intramural nodules, ascites, necrosis, or peritoneal carcinomatosis, and the source organ (ovary, pancreas, kidney, stomach) can usually be identified. Multicystic mesothelioma is seldom diagnosed at preoperative imaging because it is exceedingly rare; the diagnosis requires histological evaluation.

The treatment of choice is complete surgical excision. Complete removal of the cystic lesion, if possible, is the best treatment and the only hope in avoiding local recurrence. Aggressive surgical approaches including cytoreductive surgery with peritonectomy are recommended. [8,12] Hormonal therapy with anti-estrogens and gonadotrophin-releasing analogues, sclerotherapy with tetracycline, hyperthermic peritoneal perfusion with cisplatin and peritonectomy with intraperitoneal chemotherapy have also been attempted in individual cases with varied degrees of success. Adjuvant chemotherapy and radiotherapy are not indicated as this tumour has a prevailing benign character.[8,12]

The prognosis is excellent and the only death that has been reported in the literature occurred in a patient who refused to undergo resection 12 years after diagnosis. [4] A 36-year-old woman was followed for 10 years, and developed an aggressive, diffuse malignant mesothelioma after six surgical procedures. [13]

CONCLUSION

Benign multicystic peritoneal mesothelioma is a very rare benign cystic tumour. This lesion has a non-specific appearance on imaging which does not permit differential diagnosis from other cystic lesions and always requires histological evaluation. It has a high recurrence rate after surgical resection but malignant transformation has very rarely been reported. A systematic follow-up of these patients is required and further resection or other therapy may be indicated. Although approximately 130 cases have been described in the literature, publication of case reports on BPM is required to better define this entity.

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