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Case Report

Benign Cystic Mesothelioma- Masquerading as Mesenteric Cyst

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ABSTRACT

We report a case of benign cystic mesothelioma which clinically and radiologically appeared to be a mesenteric cyst but histopathology showed it to be a benign cystic mesothelioma. We also noted it is an uncommon condition in males. It is a rare condition that arises from abdominal peritoneum. Benign cystic mesothelioma usually affects premenopausal women and is extremely rare in men. Many factors such as insult to peritoneum like previous abdominal surgery, endometriosis, PID are suspected to contribute to its development. The main management is complete surgical excision but as in this case it was adherent to vital structures only 80% of cyst could be excised. Malignant transformation is rare.

Keywords: benign cystic mesothelioma, peritoneal inclusion cyst, BCM

INTRODUCTION

Benign cystic mesothelioma (BCM) is rare disease characterized by proliferative lesions of the peritoneal mesothelial cells mainly in the pelvic and abdominal cavity. However it can also develop in pleura, pericardium, tunica vaginalis and spermatic cord. [1] In the past it has also been called as peritoneal mesothelioma, Multicystic multilocular cyst of peritoneum, multicystic mesothelioma and multicystic peritoneal inclusion cyst. [1,2] It was first described by Mennemeyer and Smith (1979) [3] as a possible diagnosis in a case that resembled intraabdominal cyst hygroma. Many factors such as insult to peritoneum like previous abdominal surgery, endometriosis, PID are suspected to contribute to its development. The rate of BCM in men is 17%. [4]

CASE PRESENTATION

A 65 yrs old male patient came to OPD with complaints of swelling on the left side of abdomen since 3 months. Patient had no other complaints. On per abdomen examination a large lump approximately 12*8 cms, cystic in consistency with dull note on percussion was noted. Lump was partially mobile. No previous surgical scar was seen on the abdomen. Rest of the abdomen was soft. USG was suggestive of mesenteric cyst or a duplication cyst of the bowel. CECT abdomen and pelvis was performed which showed large simple cyst probably Retroperitoneal in left upper quadrant of abdomen extending upto L4-L5 level with one of the jejunum coils stretching around the anterior wall of the cyst. It also showed mildly dilated left pc system due to compression.

All routine blood investigations were normal. Exploratory laparotomy was planned followed by excision. Intra

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Operative Findings-Large Cystic Swelling Retroperitoneal (Figure 1) with loop of jejunum around anterior wall of cyst was noted. An attempt was made to excise the cyst but as it was adherent to vital structures like renal pelvis and large vessels, decision was taken to partially excise the cyst wall (upto 80%). Drain was kept and monitored. Patient was discharged on 14th Post op Day with full recovery. Patient visited for follow up after 15 days. Histopathology report was suggestive of benign cystic mesothelioma. (Figure 2)

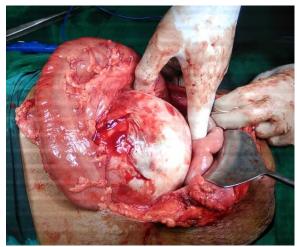


Figure1 showing Large cyst in Left upper quadrant

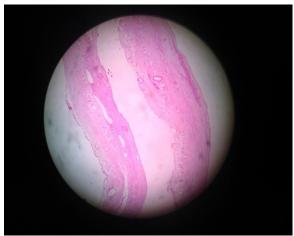


Figure 2 Cyst wall lined focally by mesothelial cells. The wall is composed of fibromuscular tissue, many congested blood vessels, edema and sparse mononuclear cell infiltrate.

DISCUSSION

Although BCM was first referred to by Plaut ^[5] in 1928, it was suggested to have its origin in the mesothelial cells due to the observations made by Mennemeyer and Smith ^[3] using electron microscopy. Later

the disease could be diagnosed as soon as it had been distinguished from other diseases. After the introduction of immunohistochemistry, BCM was diagnosed as it stained both for calretinin and D2-49 at the same time.

It is known to be more frequent in patients with a history of endometriosis, pelvic inflammation, streamlined treatment plan. Currently aggressive surgical resection is the mainstay of treatment with palliative debulking and reoperation for recurrence (15,11,5). With up to 50 percent recurrence rates and its malignant potential, debulking surgery does not appear to be the most acceptable treatment option for these patients. Patients may suffer from poorly controlled chronic abdominal and pelvic pain (15). Uncertain results have been seen after abdominal surgical treatment, particularly in patients having hysterectomy and cesarean section. [4] BCM is a tumor originating from mesothelial cells in the peritoneum, pleura and pericardium. It can also appear in the groin and scrotum, which is the extension of the peritoneum. In the case of BCM in the groin and scrotum, the patients can detect the palpable masses by themselves and therefore present themselves to the hospital earlier. [1] On the other hand, BCM in the abdominal or pelvic cavity is not easy to be diagnosed unless the mass is large. BCM in this case was also not easily detected as it is rare in a middle-aged man.

BCM can be classified into three types depending on its shape: (1) the solitary type when the boundary is clear, (2) the localized type when multiple masses are found in a localized part of the abdominal membrane, and (3) the diffuse type where the masses are widely spread across the abdominal membrane. [4] Surgical removal is the best treatment. To prevent possible recurrence and transformation malignancy, it is recommended to cut out a wide enough area whenever possible. [6] Worldwide, there were only 2 cases where BCM developed into malignancy: a baby at the age of 6 months and a female at the age

of 36 years. ^[7,8] Accordingly, as the rate of transformation to malignancy is very low, re-excision is often performed even for local recurrence. In a few studies, there have been attempts to use heated intraperitoneal chemotherapy in the abdominal cavity, but this is still in an experimental stage. ^[9]

Due to its rarity, BCM treatment options remain an area of controversy and there is no streamlined treatment plan. Currently aggressive surgical resection is the mainstay of treatment with palliative debulking and reoperation for recurrence. [6,8,10] With up to 50 percent recurrence rate and its malignant potential, debulking surgery does not appear to be the most acceptable treatment option for these patients. Patients may suffer from poorly controlled chronic abdominal and pelvic pain. [10] Uncertain results have been reported with patients receiving adjuvant chemotherapy and/or radiation therapy. [8] Other approaches such as sclerotherapy with tetracycline, continuous hyperthermic peritoneal perfusion with cisplatin, and antiestrogenic drugs have been suggested. [6] The optimal treatment may be cytoreductive surgery with peritonectomy combined with perioperative intraperitoneal chemotherapy to eliminate all gross and microscopic disease. [8] The goal of this treatment regimen is to reduce the likelihood of progression or recurrence.

CONCLUSION

BCM is a very rare disease, which is often observed in women in their pre menopausal age, complete excision is the gold standard treatment for benign cystic mesothelioma, however incomplete excision will lead to recurrence and hence regular follow-up is required.

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