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Benign cystic Mesothelioma of Peritoneum: A Case Report and Review of Literature

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Abstract:

Benign cystic peritoneal mesothelioma (BCPM) is an uncommon clinical condition with diverse presentations. Its abstruse pathogenesis and association with diverse intra- abdominal conditions make an explicit diagnosis arduous. Diagnostic veracity and conscientious follow-up are essential because, though benign nature, it recurs locally with potentials for malignant transformation. Hence a definitive diagnostic and surgical strategy needs to be formulated.

Key words: *Cystic lesion, Mesothelioma, malignant transformation, Peritoneum, Surgery*

Introduction:

Benign cystic peritoneal mesothelioma (BCMP) is a rare benign neoplasm arising from the epithelial and mesenchymal elements of the mesothelial cells. It was first reported by Mennemeyer and Smith in 1979 and only about 130 cases have been reported in the literature worldwide. The lesion frequently occurs in women during their reproductive years[3] and is commonly associated with a history of previous abdominal surgery[4], endometriosis[5] or pelvic inflammatory disease[5]. It is known to occur in children[6], man[7], and at other extra abdominal sites[8,9]. The high recurrence rate[10], potential for malignant transformation and varied clinical presentation warrants definitive surgical guide line.

This is a case report of 44 year female patient, who presented with diffuse abdominal pain and abdominal distension. The clinical and radiological findings suggested ovarian neoplasm but histological evaluation of the specimen extracted after the laparotomy confirmed it to be benign cystic mesothelioma of peritoneum.

Case Report:

A- 44- year old woman presented to Gynaecological Oncology Out Patient Department of Delhi State Cancer Institute, Delhi; with diffuse abdominal pain, loss of weight, decreased appetite and abdominal distension. Patient was post menopausal for three years, with gravida four and no family history of any malignancy. Past medical history was unremarkable. She gave a history of bilateral tubal ligation eight years earlier. Abdominal examination revealed an ill defined tender abdomino-pelvic mass of around 18-20 wks pregnant uterus size. It was ill defined intra-abdominal mass in the right. iliac fossa extending to midline and firm in consistency with almost regular surface but the margins could not be defined. There was no evidence of clinical ascites or any other organomegaly. Pelvic examination corroborated the findings of abdominal examination with a fixed, almost immobile, firm and tender mass extending in pouch of Douglas (POD).There was no deposits in POD. Patient was afebrile with normal vital signs. Abdominal ultrasound reported a well defined septated cystic lesion of 59x39 mm in size in right adenexa with small amount of fluid around the cystic mass and in the pelvis.

PETCECT reported fluorodeoxyglucose (FDG)avid mass lesion in right adnexa with diffusely increased FDG uptake along peritoneum and omentum. CA 125 and other tumour markers (Ca19.9, CA72.4, CEA, alpha fetoprotein and LDH) were normal. She was diagnosed having ovarian malignancy and exploratory laparotomy performed.

Surgical findings revealed left complex solid adnexal masses of 4×5 cm and whole peritoneal cavity studded with gelatinous fluid filled vesicles. Multiple free intraperitoneal cystic lesions of various size were observed floating in the peritoneal cavity. Since frozen section facility was not available at that time, the mass and few cysts were harvested and extracted for histological examination. The pathology report was benign cystic mesothelioma of peritoneum and ovary. Her postoperative period was uneventful. During the follow up, she was found to have recurrence at 6 weeks

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in the form of 5×6 cm cystic mass in left iliac fossa without any clinical complaint.This mass was non FDG avid on PETCECT.

Patient was put to surgery again with complete cytoreduction with hysterectomy.Since the pelvic lymph nodes were enlarged and palpable, bilateral pelvic lymph node dissection was done. She has been on close follow up now for the last three years without any complaint or clinical findings and is leading good quality of life.

Discussion:

Mesotheliomas are mesenchymal neoplasm originating from the serous lining of the pleural, pericardial or peritoneal space. In most of the cases it arises from the pelvic surfaces of the peritoneum and is benign in nature. The etiology remains unclear, but it is well known that many inciting factors may promote hyperplastic and neoplastic changes in mesothelial cells. The suggested provoking factors are foreign fibers and dusts, inflammatory mediators, and mechanical injuries[6]. Proliferation and inward migration of peripheral mesothelial cells, proliferation and metaplasia of underlying connective tissue cells, and surface attachment and differentiation of free-floating mononuclear cells all have been postulated as the mechanism of mesothelial cell proliferation in pathological conditions.[6]The pathogenesis of BCM is still unclear, however as the majority of cases occur in females who are in reproductive age group , it is believed that female sex hormones play a role.[7]

The most common presenting symptoms are chronic or intermittent abdominal or pelvic pain, abdominal distension with pelvic mass. Preoperative diagnosis of BCM is challenging often it can only be made at the time of surgery. Imaging modalities that can be used include abdominal ultrasound where it is appears as multicystic, vascular mass without calcifications[8]. CT scan /MRI is useful to visualize the lesion, but is not helpful in differentiating it from other cystic lesions of the peritoneum especially lymphangiomas, pseudomyxoma peritonei and endometriosis.[9]

Aspiration cytology can be useful in making a preoperative diagnosis[10]. Laparoscopy remains the best diagnostic tool because it enables to perform biopsies and to establish the diagnosis.[11] Definitive diagnosis depends on histological examination demonstrating multiple grapelike clusters of mesothelium-lined cysts, which may be unilocular or septate .

Because of its benign nature, adjuvant chemotherapy and/or radiotherapy are not indicated for patients with BCMP. Several authors have demonstrated the presence of estrogen and progesterone receptors in normal peritoneum and have proposed the use of gonadotrophin releasing hormone agonists as therapy.[12,13]

Some researchers have advocated aggressive surgery (extended peritonectomy) followed by heated intraperitoneal chemotherapy (HIPEC).[14]

Though there are chances of recurrence, recommended treatment of choice is aggressive surgical exploration with cytoreduction and peritonectomy.[15]

Periodic follow-up for at least once in 3 months for first 2 years, 6 months in next 2 years and yearly thereafter is needed. Further studies are needed to better understand its etiology and pathogenesis.

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There is no conflict of interest and or financial obligations to anyone and due consent has been obtained from the patient and the organization for publication of this manuscript.

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