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Case Report

Benign multicystic mesothelioma with ovarian endometriosis: a rare case report with review of literature

Basanta Manjari Hota*, Kavitha Bakshi, Naimisha Movva, Swathi Pandirla

Department of Obstetrics and Gynecology, Mamata Medical College, Khammam, Telangana, India

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***Correspondence:**
Dr. Basanta Manjari Hota,
E-mail: drmanjarihota@gmail.com

ABSTRACT

Benign multicystic mesothelioma is a rare pathology that is usually diagnosed on laparotomy for other abdominal diseases. It is more common in females of reproductive age group than in males. The commonest presentation is chronic pain abdomen. Ultrasound is the usual diagnostic tool and the ovarian cyst is the commonest differential diagnosis. Both, reactive and neoplastic hypotheses are considered in the etiopathogenesis of this condition. Management of the disease constitutes complete surgical excision and follow up of the patient. Our patient, a 33 years old parous lady, reported with chronic pain abdomen and right adnexal multiloculated cystic mass. She underwent laparotomy with a provisional diagnosis of ovarian cyst. Histopathology of the excised specimen was benign multicystic mesothelioma with ovarian endometriosis. This case was presented for its rare occurrence, mimicking malignancy, the chance of local recurrence and malignant transformation requiring long term follow up.

Keywords: Mesothelioma, Benign, Endometriosis, Neoplastic, Reactive

INTRODUCTION

Mesothelioma is a proliferative neoplasm consisting of epithelial and mesenchymal cells of mesothelium which form serosal covering and lining of various organs in the body. It can be benign or malignant. Benign multicystic mesothelioma is a rare entity. It is often diffuse and shows a marked predilection for the surface of pelvic viscera like ovary, uterus, bladder and rectum.[1] Extra pelvic sites of origin are pleura, pericardium and peritoneum. This is a rare disease with challenges in determining its origin, pathogenesis, diagnosis and treatment. Here, we presented a rare case of benign multicystic mesothelioma in a 33 years old lady, who presented with a complex ovarian cyst.

CASE REPORT

A 33 years old lady, with a history of two normal deliveries and tubectomised 11 years back, reported to the outpatient department of obstetrics and gynecology in Mamata Medical College, Khammam, Telangana with pain abdomen of 6 months duration. Pain was in right iliac fossa, intermittent, dull aching and insidious in onset. There was no history of abnormal vaginal discharge, dyspareunia, fever or abnormal bowel or bladder habit. She was a non-smoker. There was no other contributing history. Her general and systemic examination was within normal limits. Thyroid and breasts appeared normal. The abdomen was not distended and nontender. There was no palpable mass and free fluid. The tubectomy scar was healthy. Vaginal bimanual examination revealed uterus of normal size, firm, mobile and non-tender. Left fornix was free and nontender. The right fornix had some irregular non-tender mass with variegated consistency. With the provisional diagnosis of right adnexal mass, the patient was investigated. Her complete blood picture, urinalysis, biochemical parameters, PAP smear, serum CA-125, viral markers and X-ray chest were within normal limits. Ultrasonography (USG) showed right adnexal multilobulated cystic lesion of 8.9×3 cm with minimal free fluid in peritoneal cavity. With the provisional diagnosis of right ovarian cyst, patient was posted for surgery.

Laparotomy revealed peritoneal cavity studded with multiple cysts of various sizes containing clear fluid, arising from peritoneum, omentum and serosa of the gut, uterus and right ovary (Figure 1). The largest cyst was 1×1 cm in size. The left ovary, fallopian tube and uterus appeared normal. Right salpingo-oophorectomy, omentectomy and complete excision of the cystic lesions were performed (Figure 2). There was no palpable lymph node. The liver and under surface of diaphragm appeared normal. The postoperative period was uneventful. Histopathology report of the right ovary revealed features of ovarian endometriosis (Figure 3). Histopathological study of the cysts showed features of benign multicystic mesothelioma associated with focal mesothelial hyperplasia (Figure 4).The patient was discharged on 10th postoperative day after an intramuscular injection of depomedroxy progesterone acetate (150 mg) with advice for review after 3 months. However, she reported for review after 9 months, when she was found to be asymptomatic except for menorrhagia in two previous cycles. Clinical examination revealed no abnormality. Hemoglobin was 8.0 gm%. USG abdomen showed 5-6 tiny thin walled anechoic cysts in the right iliac fossa, the largest one being 8×5 mm. The uterus and left ovary were normal. She was treated with iron and folic acid, injection depomedroxy progesterone acetate 150 mg and advised periodic review.

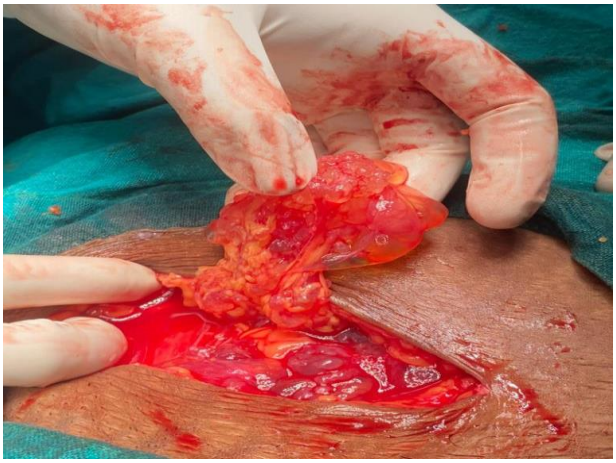

Figure 1: Per op findings of multiple cysts with omentum.

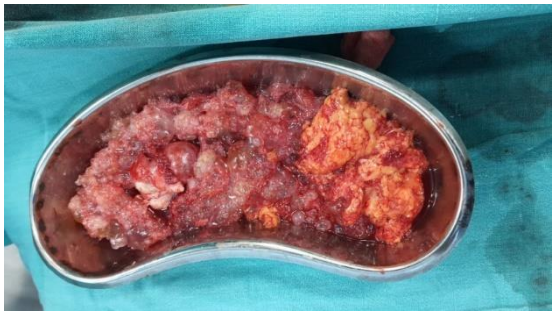

Figure 2: Resected specimen of cysts along with omentum and right ovary.

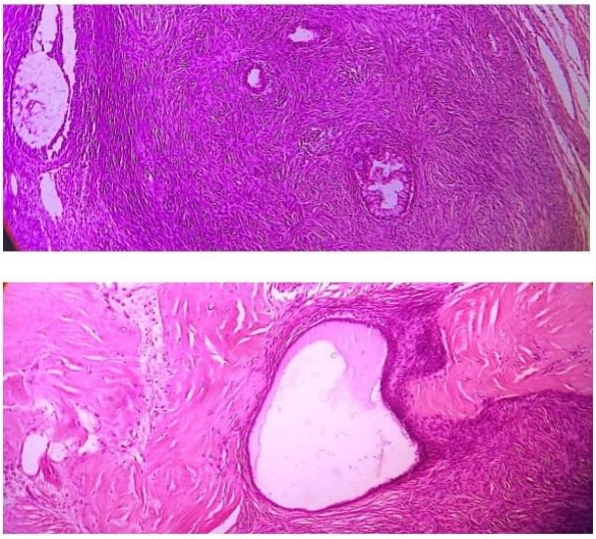

Figure 3: Sections of right ovary showing extensive areas of fibrosis, necrosis and cystic changes with scanty endometrial stroma.

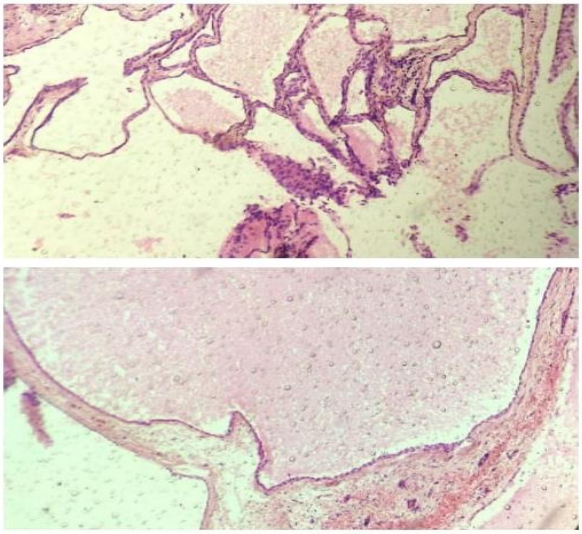

Figure 4: Sections of cyst membranes showing flattened to cuboidal mesothelial cells (calretinin +ve) with luminal eosinophilic proteinous material and focal mesothelial proliferation.

DISCUSSION

Benign multicystic mesothelioma is a rare disease and was first described by Plaut in 1928, whereas its mesothelial nature was discovered by Mennemeyer et al in 1979.[2] Only 200 cases have been reported till 2017 with 83% being female.[3] It is common among women in 20-40 years of age. The commonest presentation is chronic or intermittent lower abdominal or pelvic pain, tenderness or distension with an abdominal or pelvic mass. Our patient was 33 years old with chronic pain abdomen. The etiology of benign mesothelioma was not very clear like malignant mesothelioma due to asbestos exposure.[1] Pathogenesis of

benign multicystic peritoneal mesothelioma was not clearly known and there was some controversy regarding its neoplastic and reactive nature.[1,4] Hyperplasia of mesothelial cells in response to various stimuli like foreign material, dust, small fibers, trauma or mechanical injury may lead to over proliferation of mesothelial cells and metaplasia of underlying tissue producing the pathologic lesion. Women with this lesion often have a history of prior pelvic surgery, endometriosis or pelvic inflammatory disease.[5] Our patient had history of abdominal tubal sterilization and ovarian endometriosis was confirmed on histopathology. Social risk factors include alcohol and smoking. Family history of the pathology may be associated.[5,6] Diagnosis is usually incidental. In the present case, laparotomy was done with provisional diagnosis of ovarian cyst and chronic pain abdomen. Management of the disease constitutes complete surgical excision of the lesion and follow up. In our case, detection of a few tiny cysts in the right adnexa on follow-up USG at nine months of surgery might be due to residual lesion or recurrence. The patient was advised for further follow up and progesterone treatment. The prognosis of this benign condition is uncertain. Recurrence after the initial management and malignant transformation though rare has been reported.[1,7,8] Thus, routine follow up and imaging are required postoperatively for the lifetime.[2,9] Recurrent cases may need repeat surgery and hormone therapy like anti-estrogen or Gonadotropin agonist.[10,11] As our patient had associated ovarian endometriosis, she was offered long acting progesterone treatment.

CONCLUSION

Benign multicystic mesothelioma is not only a rare pathology but also is challenging with its diagnosis and management. Complete resection of the lesion is the primary treatment of choice. As there is a chance of local recurrence and malignant transformation, lifetime follow up of the patient is recommended.

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