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Case Report

Benign multicystic mesothelioma of peritoneum presenting as a tubo-ovarian mass – A case report

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Multicystic mesothelioma is a rare benign neoplasm of the peritoneum occurring in women of reproductive age. A 45 year old woman presented with a mass per abdomen measuring 20 x 18 cms^2 with on and off pain since 5 months. She had a history of five abdominal surgeries in the past. USG and CT scan of the pelvis showed a multicystic mass of 20 x20 cms^2 arising from right adenexal region and a provisional diagnosis of ovarian cystadenocarcinoma was suggested. A total abdominal hysterectomy and bilateral salpingo-oophorectomy was done. The specimen showed a multilocular cystic mass of 20 x 15 cms^2 adherent to the uterus and cervix in the right adenexal area. Histology showed cystic spaces lined by cuboidal cells with hobnail appearance. This case is presented for its rarity and its relationship with history of previous surgical interventions in abdominal region.

Keywords: Multicystic mesothelioma, benign mesothelioma of peritoneum, benign multicystic mesothelioma

INTRODUCTION

Benign multicystic mesothelioma of the peritoneum (BMPM) is a rare lesion arising from peritoneal mesothelium that covers the abdominal and pelvic cavity. It occurs frequently in women during their reproductive years and is associated with a history of previous abdominal surgery, endometriosis or pelvic inflammatory disease (Michael et al., 2006). This lesion was first described by Menemeyer and Smith in the year 1979 (Mennemeyer and Smith,1979).There are reports of this lesion occurring in children, in men and rarely in exta-abdominal regions (Michael et al., 2006). As per the review of the literature approx 130 cases have been reported (Michael et al., 2006; Weiss and Tavassoli, 1988). The origin of this rare lesion is known, but the pathogenesis is not clear. We report a case of this rare lesion in a middle aged woman who was admitted in the ward of Obstetrics and Gynecology with a pelvic mass.

Clinical History

A 45-year woman was admitted in the ward of Obstetrics and Gynecology with complaints of mass per abdomen of 5 months in duration and gradually increasing in size, with on and off pain in abdomen. The mass was restricting her routine work and she had urinary incontinence for 3 months, but no bowel disturbances.

She had a history of 5 surgeries in the past. The 1st operation was a laparotomy done for repair of an accidental injury to the abdomen 20 years back, followed 6 months later by 2nd laparotomy for tubectomy. The third surgery was done 4 years later for release of post-operative intra-abdominal adhesions. This was followed by fourth surgery 2 years later by another laparotomy for drainage of pelvic abscess. The fifth surgery was 1 year back for right ovarian cyst.

On clinical examination, a huge mass was felt in the lower abdomen and pelvis measuring about 20x15 cms^2, soft to firm in consistency. A provisional diagnosis of a large ovarian tumour was made. An USG and CT scan of the abdomen and pelvis showed a well defined multiseptate cystic pelvoabdominal mass measuring 20x18 cms^2 arising from the right adenexal region.The uterus was not seen clearly, probably compressed to the left lateral pelvic wall. The lesion showed a focal nodular enhancing lesion on the posterolateral aspect.There was no evidence of omental deposit or fluid in the abdomen. Radiologically a diagnosis of a large right ovarian cystic mass with focal enhancing area, possibly an ovarian cystadenocarcinoma was suggested. The patient had no toxic signs. Her routine blood counts and biochemical parameters were within normal limits. The tests for CA125 and CA19-9 were not done.

The patient underwent a total abdominal hysterectomy with salpingo-ophorectomy with removal of mass. The hysterectomy specimen was sent to department of pathology for histopathological analysis.

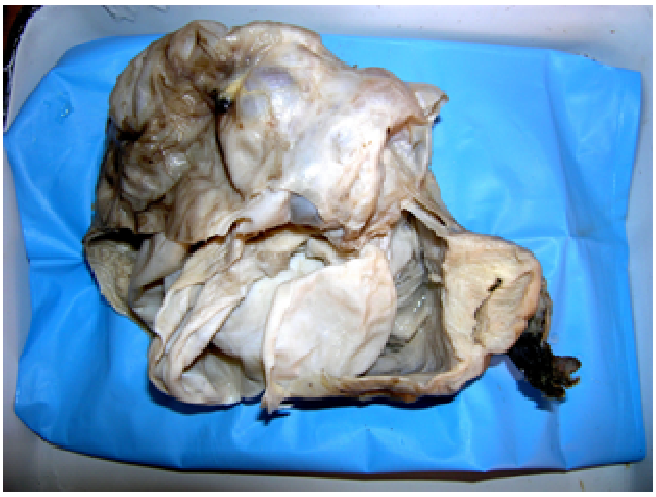

Figure 1. Gross photograph showing multilocular cyst adherent to the wall of uterus and cervix on right side.

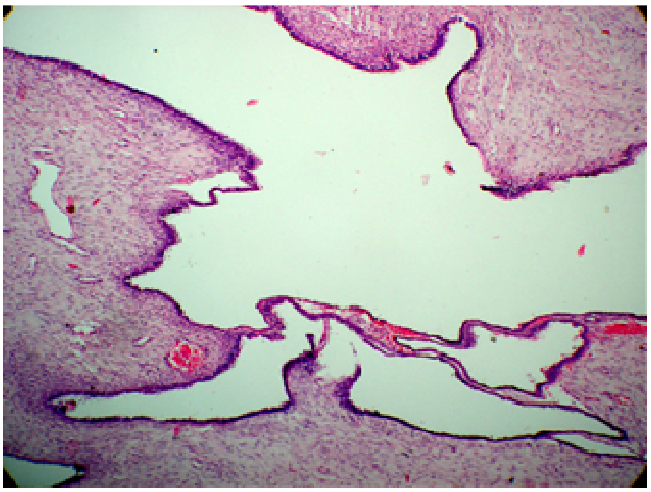

Figure 2. Photomicrograph showing cystic spaces and intervening connective tissues stroma (H&E, X10).

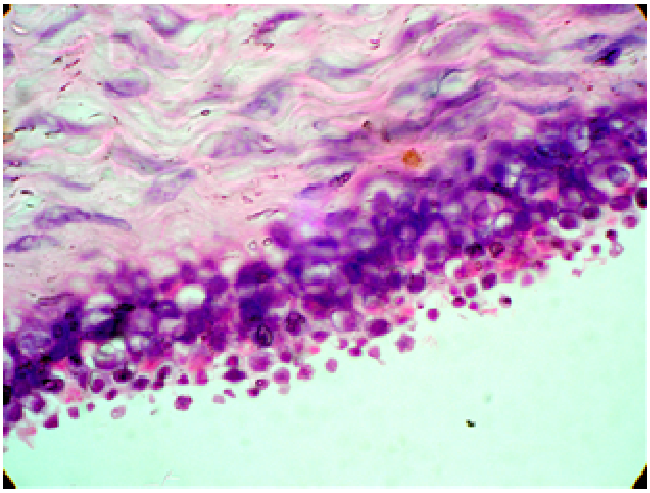

Figure 3. Photomicrograph showing cystic spaces lined by cells with hob-nail appearance (H&E, X40)

Pathological Findings

Grossly the specimen was consisted of uterus, cervix and left sided adenexa. The right sided adenexa could not be identifed. The left sided adenexa was grossly normal. A multilocular cystic structure measuring about 20 cms across was adherent to the surface of uterus and cervix on the right side (Figure 1). On cut section, endomyometrium was unremarkable. The cyst wall was multilayered and the cysts were filled wih straw-coloured fluid. No solid areas were seen wthin the cyst. Microscopically the lesion showed cystic spaces of various size and intervening connective tissue stroma (Figure 2). These cystic saces were lined by cells which varied from flat to cuboidal with multilayered hobnailed shaped appearance in some areas (Figure 3). Endometrium was in proliferative phase, myometrium unremarkable and cervix showed chronic cervicitis. A diagnosis of benign multicystic mesothelioma of peritoneum was made. The post operative period was uneventful. The patient has not reported with recurrence in the follow-up examination for about 3 years.

DISCUSSION

Benign multicystic mesothelioma is a localized tumor arising from epithelial and mesenchymal elements of the peritoneum and does not metastasize. It has a strong predilection for the surface of pelvic viscera (Michael et al., 2006).

The pathogenesis of this benign tumour is controversial. The association of this tumor with inflammation, endometriosis and a history of prior surgery, suggests that multicystic peritoneal mesothelioma is probably a peculiar peritoneal reaction to chronic irritation stimuli with mesothelial proliferation and cyst formation (Groisman and Kerner, 1992; Michael et al., 2006; Tangitamol et al., 2005 , Weiss and Tavassoli, 1988)

The differential diagnosis of this lesion can be a number of benign and malignant lesions that can present a cystic mass, such as lymphangioma, endometriosis, cystic adenomatoid tumor, cystic forms of endosalpngiosis, Ovarian cystadenoma, Ovarian cystadenocarcinoma, pseudomyxoma peritonei, necrotic leiomyoma or leiomyosacoma (Michael et al., 2006; Weiss and Tavassoli,1988). Cystic lymphangioma occurs more commonly in male and is restricted to mesentry, omentum, mesocolon, and retroperitoneum but rarely in ovary. On gross examination, the cystic component is often chylous and microscopic examination reveals smooth muscle and lymphoid tissue (Michael et al., 2006). Endometriotic cysts typically contain dark

chocolate brown materials and are composed of endometrial stroma lined by endometrial glands (Michael et al., 2006). The cystic forms of endosalpingiosis consists of cystic spaces lined by tubal type of epithelium (Michael et al., 2006). A cystic adenomatoid tumor of the uterus can come as a differential diagnosis as it simulates lymphangioma grossly and composed of multiple cystic spaces lined with cuboidal cells like mulicystic mesothelioma but the stroma of cystic spaces contain smooth muscles which differentiates it from multicystic mesothelioma (Isac et al., 2008).

The malignant conditions which mimic benign multicystic mesothelioma will have features of malignancy like cellular atypia, increased mitotic counts and stromal infiltration.

Although with the available imaging techniques, this lesion can be demonstrated but Benign Multicystic mesothelioma is seldom diagnosed at pre-operative imaging because it is exceedingly rare; the diagnosis requires histologic examination. However a differential diagnosis is difficult to be made from other cystic neoplastic or inflammatory lesions arising from this anatomical area. The differential diagnosis of BMPM from the cystic tumors of the ovaries is important since BMPM may be treated by local excision with preservation of the ovaries. Although there are no efficient tools for an early diagnosis, a long term follow-up is needed due to high incidence of multiple recurrences after surgical resection (Philip et al., 2004; Weiss and Tavassoli, 1988). This tumor does not have a tendency to transform into malignancy (Philip et al., 2004)

Conclusion

Benign multicystic mesothelioma can be considered as a pre-operative differential diagnosis in a woman presenting as tubo-ovarian mass with a history of previous abdominal surgeries. A regular follow up for long term is a must even after surgery because of its high incidence of recurrence.

Although, mesothelioma is a rare tumor, it is important for all gynecologists to recognize its existence, the appearance of this lesion and its generally benign course.

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