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Case Report

Endometrioid adenofibroma of ovary-a two faced tumour: case report

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ABSTRACT

Endometrioid variant of the adenofibromas accounts for only 1% of epithelial neoplasms of ovary. Though benign, specimen of endometroid adenofibroma of ovary needs to be evaluated by an experienced pathologist to rule out borderline and malignant cases. This prevents unnecessary adjuvant therapy for benign cases. We report a case of a large endometrioid adenofibroma arising at the left ovarian fossa in a post-menopausal woman who had undergone total abdominal hysterectomy with bilateral salpingo-oophorectomy 6 years back. Recurrence of endometroid adenofibroma of ovary is rare, however, long term follow-up is mandatory due to its low malignant potential. Though benign, specimen of endometroid adenofibroma of ovary needs to be evaluated by an experienced pathologist so that borderline and malignancy can be ruled out. Endometroid adenofibroma of ovary though benign needs long term follow up due to its low malignant potential.

Keywords: Ovary, Endometriod adenofibroma, Recurrence, Benign

INTRODUCTION

Ober, in 1959 first proposed the term "adenofibroma" to a rare benign Mullerian mixed tumor.[1] Endometrioid variant of this adenofibromas accounts for only 1% of epithelial neoplasms of ovary, of which 83% are unilateral.[2] Endometrioid adenofibromas (EA) are commonly found in the post-menopausal age group. We report a case of a large EA arising at left ovarian fossa in a post-menopausal woman who had undergone total abdominal hysterectomy with bilateral salpingo-oophorectomy (TAH BSO) for benign ovarian cyst 6 years back.

CASE REPORT

A 66-year-old woman came to the out patient department (OPD) of obstetrics and gynaecology of central referral hospital, Sikkim, with history of pain lower abdomen for one month and a ultrasonography report showing complex large mass in pelvis (13 cm). She had undergone TAH BSO for a right ovarian complex cyst (10 cm), 6 years back in a hospital outside Sikkim. Histopathology of the uterus with cervix was normal, however, ovary showed dysplasia (side not mentioned). She came to our OPD after 6 years with the ultrasound findings. On abdominal examination there was a longitudinal midline infra umbilical scar and a 28-week pregnant uterine size abdominopelvic mass which was immobile. Clinically there was no ascites and no lymphadenopathy. Vaginal vault was free of any growth; however, a mass was palpable which was fixed and high up in the pelvis. Bimanual examination confirmed the same. Contrast enhanced computed tomography (CECT) showed a large space occupying lesion (14.5 cm) in the pelvis with irregular septa showing enhancing peripheral wall suggestive of large ovarian cyst/ uterine stump mass. Tumour marker showed raised CA-125 (>600 U/L), while others (CEA, AFP, βHCG, LDH) were within normal limit. Vaginal vault Pap smear was negative for dysplasia or malignancy. She underwent an exploratory laparotomy where intraoperatively mesentery

of the sigmoid colon was stretched over a large left pelvic mass suggesting it to be a sigmoid colon mass. However, after opening up the left lateral pelvic wall peritoneum, a 20 cm mass was seen occupying the left ovarian fossa. The mass did not have any capsule and was vascular (Figure 1). The mass derived its vascularity from the left ovarian vessels and the sigmoid mesenteric small vessels. When the mass was enucleated from the left ovarian fossa there was oozing from the base which was controlled using sutures and cautery. Grossly the mass was solid in consistency without any capsule. Histopathology done in our institute reported it as borderline tumour. Samples were sent for re-evaluation at Tata memorial centre, Mumbai to rule out malignancy where a routine histopathology and an additional immunohistochemistry (IHC) was performed. Tumour was positive for Pax8, CK7, EMA and ER (60% tumour cells with strong intensity); while negative for WT-1, Calretinin and GATA3. Mib-1 was non-contributory (repeated twice) suggesting EA. Her post operative period was uneventful and she in on regular follow-up.

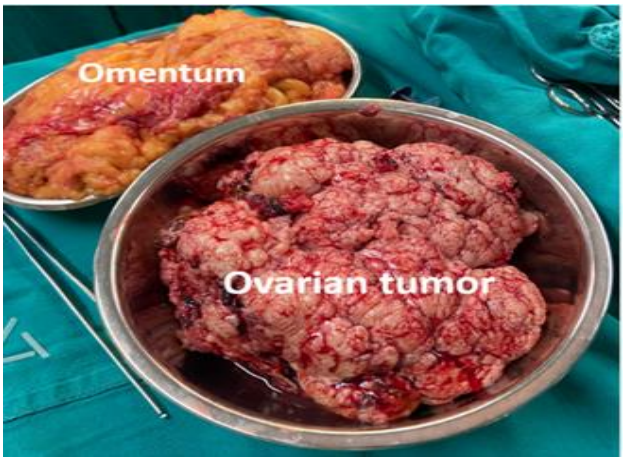

Figure 1: Specimen of solid ovarian tumour with lack of capsule along with specimen of total omentectomy.

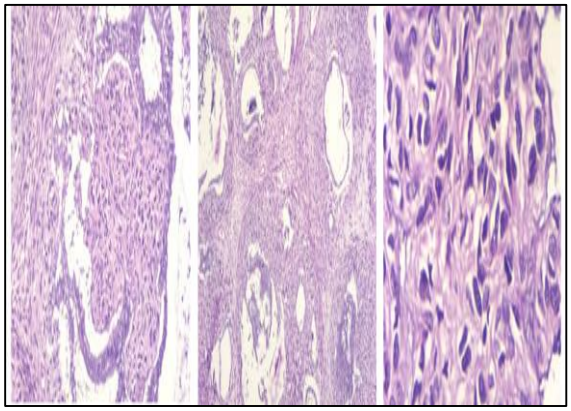

Figure 2: Histopathology of ovarian mass. 10x, H and E stain showing part of gland and stroma, glands with intervening stroma composed of spindle to epitheloid cell and 40x view show spindle to epithelial cell.

DISCUSSION

EA is an uncommon tumour which is benign and occurs commonly in the uterus. Uterine adenofibroma was first reported by Ober in 1959 when he described it as a variant of mixed mullerian tumor.[1] There are reports of this tumour arising in the cervix and the ovary. Papillary adenofibroma in the cervix was reported by Abell in 1971.[3] The tumor occurs in the post-menopausal age with the average age for occurrence being 57 years.[4] There are reports of tumor occurring in the younger age group as well.[5,6] Ovarian EA accounts for 1% of epithelial neoplasm of the ovary and 83% of these tumors are unilateral.[2] It is difficult to distinguish between borderline and benign cases. Degree of architectural and cytologic atypia helps in distinguishing between the two and thus requires a good pathologist to detect the same. DA Bell and RE Scully studied series of 27 benign ovarian masses with varying degrees of epithelial atypia in which EA was detected in 7 women.[7] These tumors had mild to severe cytologic and architectural atypia similar to that seen in atypical endometrial hyperplasia. A series of 10 cases was studied retrospectively by Roth et al.[8] They divided the cases into 2 benign, 4 proliferating, and 4 malignant tumors. They noted that proliferating adenofibromas had higher epithelial proliferation with glandular complexity and crowding compared to benign adenofibromas. Proliferating tumors may represent a form of the endometrioid tumor of borderline malignancy. The tumors in this group also frequently showed squamous metaplasia and association with endometriosis. In our patient the histology report of the ovarian mass after the hysterectomy, done 6 years back, reported as “dysplasia” which may actually have been EA. However, it is difficult to comment that the previous cyst was EA or the current mass was a recurrence, as the slides and blocks of the previous surgery was not available for review.

In our institute, the resected pelvic mass was reported as borderline tumor which was then sent to a higher oncology center (Tata memorial center) for IHC to rule out malignancy and there was high suspicion. IHC reported it as EA which may behave as well-differentiated adenosarcoma even in the absence of mitotic activity.

Ovarian EAs have fewer incidence of recurrence compared to uterine EAs. In the ovarian cases followed up for 1-18.5 years by Bell and Scully, and over 1-6 years by Rioth et al there were no incidences of recurrences.[7,8] This suggests that EA have a good prognosis. Kao and Norris, however, reported one case of ovarian EA recurrence in the vagina shortly after oophorectomy due to tumor implant, thus, raising the possibility that this tumor may have low malignant potential and will need follow-up.[9] Multiple recurrences have, however, been reported in uterine EA where conservative methods such as curettage or local excision was attempted.[10] Total surgical resection is thus the preferred treatment for EA in any site.

CONCLUSION

Ovarian EA though rare needs to be carefully evaluated as they are difficult to distinguish between borderline tumors. Lack of studies on long term follow up and possibility of these tumors possessing a low malignant potential makes it necessary for long term follow-up.

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