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Bilateral Sertoli-Leydig Cell Tumor In Postmenopausal Female - A Case Report

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ABSTRACT

Sertoli-Leydig cell tumor (SLCT) is a rare ovarian tumor that belongs to the group of sex-cord stromal tumors. These constitute less than 0.5% of ovarian tumors. [1] Most tumors are unilateral, confined to the ovaries, and are seen during the second and third decades of life. We have treated a rare case which presented in postmenopausal age group and wasinvolving both ovaries.

Keywords : Case Report, Medical, Bilateral Sertoli

Introduction

Sertoli-Leydig cell tumor (SLCT) is a rare ovarian tumor that belongs to the group of sex-cord stromal tumors. These constitute less than 0.5% of ovarian tumors. [1] Most tumors are unilateral, confined to the ovaries, and are seen during the second and third decades of life. These tumors are characterized by the presence of testicular structures that produce androgens. Hence, many patients have symptoms of virilization depending on the quantity of androgen production. The second characteristic feature of these tumors is the degree of differentiation of structures in them. The presence of these structures determines whether the tumors are benign or malignant. [2] We present the rare case report of a postmenopausal woman with bilateral SLCT. Management of these cases poses a difficult therapeutic challenge.

Case Report

59 yr old female was admitted on 24-07-08 in SKN medical college with the chief complaints of lump in abdomen since two months which was rapidly increasing in size and associated with dull pain. Patient also complained of per vaginal spotting on and off which was very minimal since two to three weeks. She was post menopausal since 12 years and had no significant menstrual complaints in the past. She had 3 children and never took any ovulation induction drugs in the past.

She was operated five years back for cancer of left breast at Sassoon General Hospitals followed by chemotherapy ; unfortunately could not produce its reports. There was no family history of ovarian or breast malignancy. She was asymptomatic in last five years after the breast surgery.

Her clinical examination revealed normal findings about vitals.Her Wt.was 58 kg with height148 cm. Right breast did not reveal any lump and Left breast was absent since mastectomy was done. There was noevidence of inguinal or cervical lymphadenopathy .Her systemic examination was normal.

On per Abdominal examination30 weeks size mass was felt arising from pelvis; lower border of which could not be reached. The mass had regular surface withrestricted mobility and wasnon tender. There was no evidence of ascites.Per speculum finding revealed an endo-cervical polyp measuring 2x2 cm which did not bleed on touch. On per vaginal examination same mass was felt is all fornices and uterus was not appreciated separately.

Investigations carried out for the surgical fitness were normal. Tumor marker CA 125 was 50.9 IU/ml. Ultrasound examination (as seen in Picture 1) revealed a large complex mass arising from pelvis extending upto epigastric region , predominantly containing multiple cysts of varying size, largest of which is 15 cm, thickened wall with echogenic projections along wall ,some are filled with echogenic material . Provisional diagnosis of Ovarian Neoplasm or mesenteric cyst was made after USG.

CT SCAN revealed 16.6cm x 22.5cm x 27 cm, large, multicystic mass arising from pelvis & reaching upto lesser curvature of stomach with multiple thin septae within, few solid components with no evidence of ascites and no evidence of metastasis.Provisional diagnosis of cyst adenoma of ovary was made after CT scan.

Her Risk malignancy index was (RMI) was calculated which was

(CA 125 x Menopausal status X USGscore)50 x 2 x 1 = 100

A differential diagnosis of Granulosa cell tumor was kept in the mind since she had spotting per vaginum.

Patient was posted for Exploratory staging laparotomy .(As seen in Picture-2).Intra- operative findings were as follow- a left ovarian tumor was seen occupying the abdomen, multicystic , with regular surface, intact capsule,without much vascularity .There was no evidence of ascites. Her right ovary, omentum, liver were within normal limits. Lymph nodes were not palpable. Hence CLINICAL STAGING was -I C. Exploratory staging laparotomy included excision of tumor with Total abdominal hysterectomy with right salphingo-ophorectomy and Infracolic omentectomy with peritoneal washings which were obtained after opening the abdomen. Her post operative period was uneventful. She required two blood transfusions. Wound healed without any problem.

The specimen was sent for histopathology which was classified as per Meyer's classification. Gross examination(as seen in Picture-3) of the pathologic specimen showed that it was an ovarian mass measuring 20 cm by 15 cm by 10cm. The external surface was smooth. A cut section of the specimen revealed solid as well as cystic areas filled with clear fluid. The histopathology examination showed a tumor composed of well-formed cords, nests, and tubules of tumor cells.Few areas of tumor cells had hyper chromatic nuclei,(as seen in picture 4) a moderate amount of cytoplasm (Sertoli cells), and occasional mitosis. Interspersed between these were nests of polygonal cells with round nuclei and abundant granular cytoplasm (Leydig cells). Retiform pattern was not seen. Right ovarian biopsy and omental biopsy was negative for malignancy. The endocervical polyp had benign histopathology.

Meyer's classification:

- Type I: Well differentiated SLCT
- Type II: Intermediate SLCT
- Type III: Poorly differentiated SLCT

In this case Sertoli Leydig cell tumor- Meyer's type II was diagnosed due to few areas of undifferentiated cells with positive peritoneal washings.

After Histopathology report patient was classified as FIGO stage Ic and was referred to medical oncologist. He had advised Chemotherapy with BEP(Bleomycin +Etoposide +Cisplatin) for 4 cycles with 3 weeks interval. After first cycle; the patient declined further chemotherapy due to cost constraint. Patient was followed up for one year with USG which was normal. We lost her follow up as patient changed her address and could not be contacted after December 2009.

Discussion

Our patient had presented at 58 years of age. The majority of these patients are seen during the second and third decades of life, with the average age at diagnosis 25 years.Around 50% of cases come to clinical attention because of progressive defeminization, as was not seen in this patient.[1]. Patient had spotting which could be due to estrogen secretion unlike testosterone.

Most of these tumors are unilateral. Bilateral tumors are found only in 1.5 % cases as was seen in our patient.75 % tumors are benign and are diagnosed in stage I, so conservative surgery in a young patient is an appropriate treatment. Our patient was 58 year old and hence underwent radical surgery. There have also been case reports of successful laparoscopic management of these tumors.[8]

Though Meyer et al. described the presence of mucinous epithelium in a SLCT in 1930, the first illustration is in the case reported by McLester in 1936, who described a SLCT containing a cyst lined by non-mucinous cells and mucinous cells, including glandular cells.[5,6]our patient did not reveal any mixed component.

The most important prognostic factors in these tumors are their stage and degree of differentiation. In a review of 207 cases by Young and Scully in 1985,[1] all well-differentiated tumors were benign, whereas 11% of tumors with intermediate differentiation, 59% of tumors with poor differentiation, and 19% of those with heterologous elements were malignant. In another study of 64 patients who had intermediate or poorly differentiated SLCT, a survival rate of 92% was noted at both 5 and 10 years.[7]

Almost all tumors are multicystic as seen in our case (1).

Adjuvant chemotherapy is considered for patients who have poor prognostic factors. The malignancy rate in tumors with heterologous elements is 15% to 20%.[1] Adjuvant chemotherapy in stage I is given to those patients who have poorly differentiated SLCT or SLCT with heterologous elements or a metastatic tumor of any histologic type.[9]The patient in this report received adjuvant chemotherapy for SLCT in stage I being in Mayer's Type II group. The following chemotherapy regimens can be considered for SLCT:

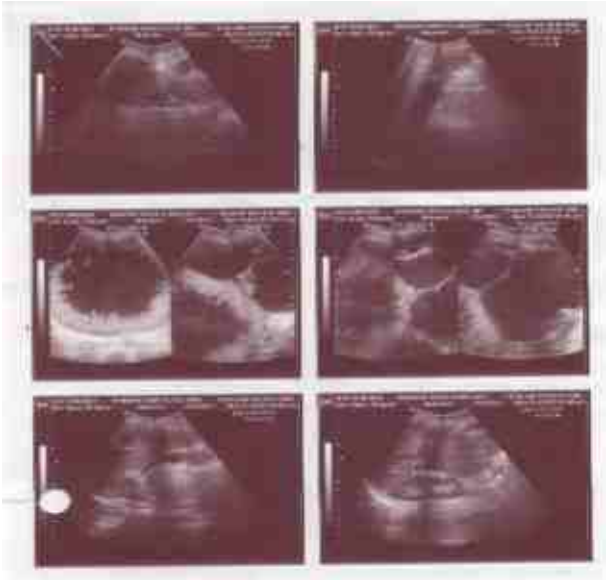
1. Cisplatin, doxorubicin, cyclophosphamide (PAC);[10]
2. Vincristine, actinomycin-D, cyclophosphamide (VAC);[11] and
3. Bleomycin, Etoposide, and Cisplatin (BEP).[12]

The BEP regimen is a comparatively safe chemotherapeutic regimen because it does not affect the fertility status of the patient.[12]

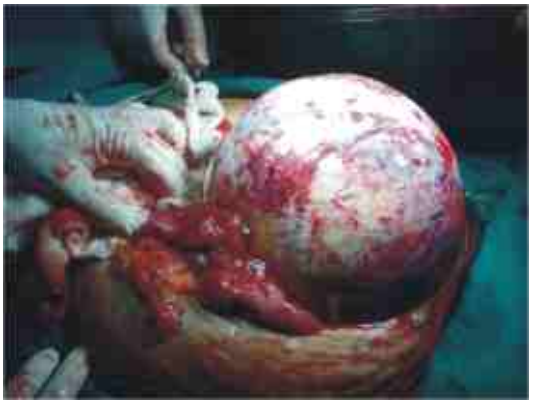
Conclusion

SLCT is a rare ovarian sex-cord tumor that usually occurs unilaterally with the mean age of presentation 25 years. However SLCT can rarely present in postmenopausal age group with bilateral ovarian mass.

PICTURE 1-Ultrasound Findings

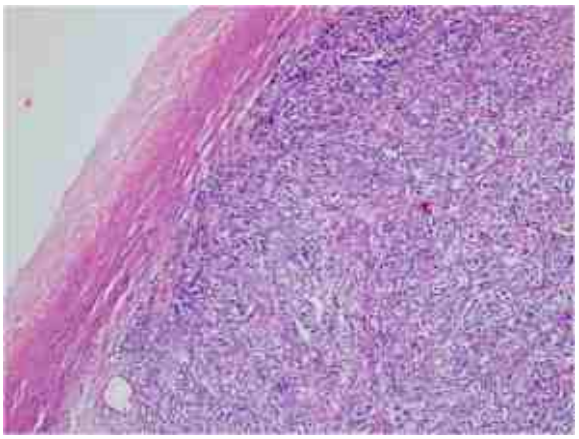


PICTURE 2- Intra- operative findings



PICTURE 3 Gross Pathology

PICTURE 4 Histopathology



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