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Case Report

Post-menopausal Sertoli-Leydig cell tumour: a report of two cases

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ABSTRACT

Ovarian Sertoli-Leydig cell tumors (SLCTs) represent less than 0.2 percent of all ovarian cancers and are an uncommon type of sex cord–stromal malignancy. Since these tumors are discovered in young women at an early stage, the management challenge is in striking the correct balance between a treatment that is effective enough to stop recurrences but yet permits fertility preservation. We present 2 case report of patients, one presented with hirsuitism and on evaluation found to have elevated levels of serum total testosterone and suspicion of SLCTs in magnetic resonance imaging (MRI) pelvis. Total laparoscopic hysterectomy (TLH) was done with bilateral salpingo-oophorectomy (BSO) and histopathology report came as ovarian Leydig cell tumour. Second case with postmenopausal bleeding and on evaluation MRI report showed right ovarian solid tumour-fibroma/fibrothecoma. Tumour markers were normal. TLH was done with BSO, histopathology report came as ovarian SLCT.

Keywords: Hyperandrogenism, Sertoli-Leydig cell tumor, Postmenopause

INTRODUCTION

Sertoli–Leydig cell tumors (SLCTs), sometimes called androblastomas, are a type of sex cord–stromal tumor that differentiate in a manner resembling that of testicles. SLCTs make up less than 0.5 percent of all primary ovarian tumors, making them uncommon.[1] The age range for this group is 2 to 75. Nonetheless, the first three decades of life account for 51% of SLCTs.[2] Extreme virilization to an asymptomatic clinical profile are just two examples of the wide range of SLCT manifestations. Since these tumors are uncommon, the imaging results are still unclear. The degree of tumor grading and staging has a substantial correlation with the prognosis of ovarian SLCTs.[3] Management of SLCT is still difficult since there are no set management protocol recommendations. For young women, fertility-sparing surgery is the best option. The course of treatment for those who have finished their family is total hysterectomy along with bilateral salpingoophorectomy.

CASE REPORTS

Case 1

A 55-year-old woman, post-menopausal since 9 years, P2L1, previous normal vaginal deliveries, with recently detected diabetes, presented with complaints of excessive hair growth since 6-7 months. It was more on the face. There was a history of hoarseness of voice and masculinisation facies. There was no history of headache or vomiting. Also, no history of post-menopausal bleeding. She presented to the endocrinology department initially.

On examination, there was hyperpigmentation and male type of hair distribution present over the face. Ferriman gallwey scoring for hirsuitism was 12. All other systemic examinations were normal. Abdominal examination and gynaecological examinations were normal.

On evaluation, her basic blood routine investigations were normal, blood sugars were elevated, with glycated

haemoglobin (HbA1c) - 6.37. Hormonal evaluation was done and serum total testosterone was elevated (738 ng/dl). All other investigations were normal, including tumor markers. Her ultrasound abdomen and pelvis revealed right ovary of size 2.9×1.9 cm with solid and cystic spaces within and increased vascularity-to rule out underlying malignancy. MRI abdomen and pelvis suggested thickened endometrium mm and heterogeneously enhancing lesion in the right ovary as shown in Figure 1. In conjunction with the clinical history needs HPE correlation to rule out SLCT. Thus, the patient referred to the gynaecology department for further management. Since the endometrium was thickened, pippelle endometrial sampling was done and the histopathology report was normal proliferative endometrium without atypia. She underwent total laparoscopic hysterectomy with bilateral salpingoophorectomy. Intraoperatively uterus was bulky for the age, right ovarian solid tumour of size 3×2 cm, left ovary normal as in Figure 2. The specimen was sent for histopathology and microscopic examination showed characteristic crystalloids of Reinke within the tumor cells as shown in Figure 3. Post-operative period was uneventful and she was discharged on third postoperative day. After one month of follow up, her serum total testosterone levels were markedly reduced to 12.1 ng/dl. Her facial features and hirsuitism was decreasing.

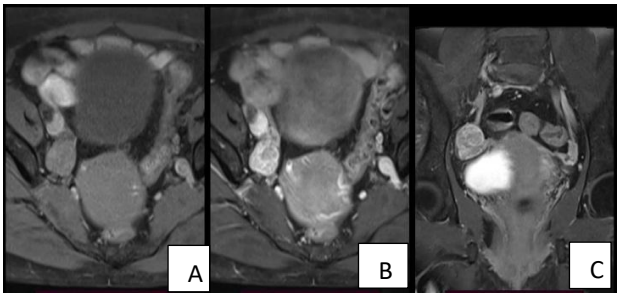

Figure 1 (A-C): Heterogeneously enhancing lesion in the right ovary.

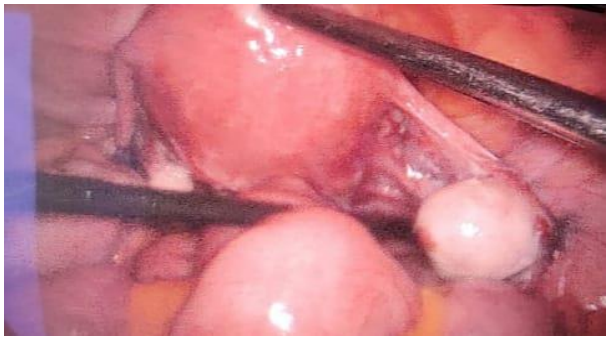

Figure 2: Intraoperative image showing right solid ovarian tumour.

Case 2

A 54-year-old para 2, live 2, previous LSCS, post-menopausal, no known co morbidities presented with complaints of post-menopausal bleeding per vagina since 1 month. She had two episodes of bleeding, each lasting for 3 to 4 days with passage of clots. Her vitals were stable and systemic examinations were normal. There was no abdominal mass palpable. Speculum examination showed atrophic cervix. Bi manual examination revealed a right adnexal mass 6×4 cm, felt through the right fornix.

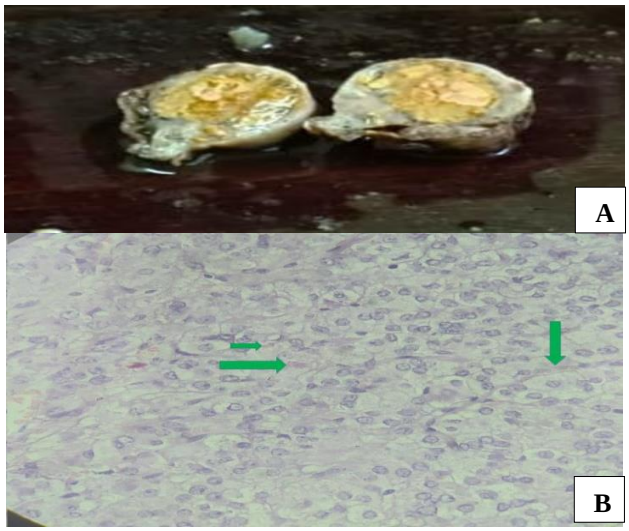

Figure 3 (A and B): Cut section of ovary with solid tumour and characteristic reinke crystals.

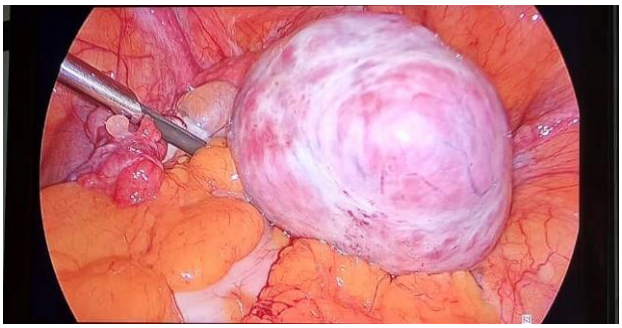

Figure 4: Right ovary with large solid mass.

Her tumour markers were normal. Ultrasound pelvis showed right adnexal hypoechoic lesion 5.9×4 cm with no cystic areas and intralesional vascularity present.

MRI pelvis showed well defined hypo enhancing solid lesion in the right adnexa, which appears uniform hypointense in T1 W and predominantly hypo intense in T2W with few irregular focal hyperintense areas possibly due to cystic degeneration. Right ovary is not well separately visualised. possible right ovarian benign lesion-fibroma/fibrothecoma (Figures 5 and 6).

Pipelle endometrial sampling was done in view of prolonged post-menopausal bleeding and histo pathological report showed endometrium with weakly proliferative glands. She underwent total laparoscopic hysterectomy and bilateral salpingoophorectomy. Intraoperatively uterus was bulky for the age, a right

ovarian tumour of size 6×5 cm, predominantly solid component (Figure 4). Left ovary a clear cyst of size 3×2 cm. Post-operative period was uneventful and she was discharged on day 3 and the histo pathological report came as SLCT- grade i well differentiated (Figures 7-9).

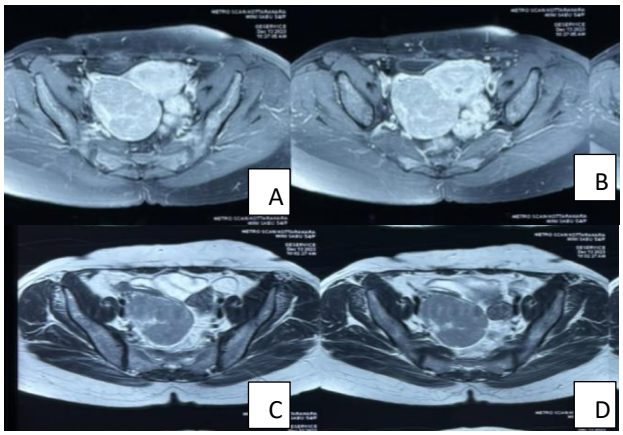

Figure 5 (A-D): Axial image of MRI.

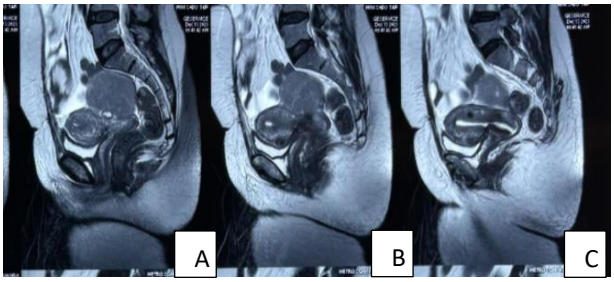

Figure 6 (A-C): Hypo intense with few irregular focal hyperintense areas.

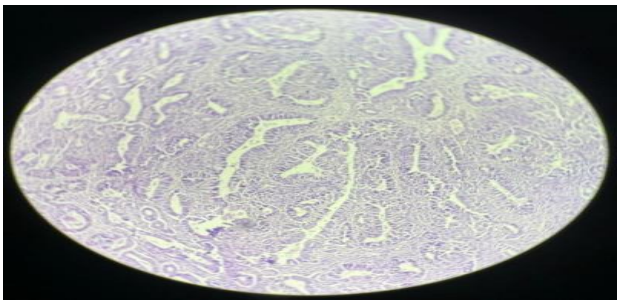

Figure 7: Characteristic Sertoli Leydig cells.

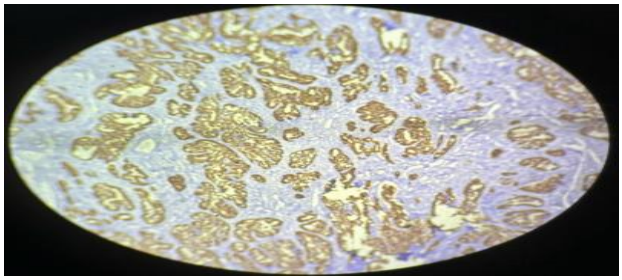

Figure 8: Immuno histochemistry showing cytokeratin positive Sertoli cells.

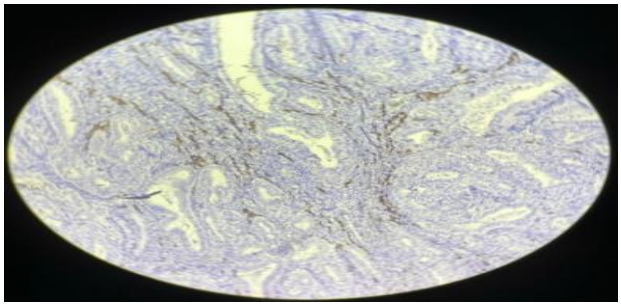

Figure 9: Immuno histochemistry showing calretinine positive Leydig cells.

DISCUSSION

Less than 0.5 percent of primary ovarian neoplasms are SLCTs, an uncommon and diverse class of ovarian neoplasms that fall within the sex cord-stromal category of cancers.[4] Any age group can experience ovarian SLCT, however young women in their 25s and 28s are more likely to experience it. Less than 10% of ovarian SLCT happens before menarche or after menopause.[5] In the category of ovarian SLCTs, which are generally uncommon, pure Sertoli cell tumors (SCTs) account for 4% of tumors and are less prevalent than SLCTs. Leydig and Sertoli cell tumors are infrequently estrogenic, generate testosterone, and can occasionally present with hirsutism, acne, deepening of the voice, clitorial hypertrophy, amenorrhea, or irregular menstrual cycles occasionally in addition to pelvic masses.[7] The most prevalent form is virilization. Secondary amenorrhea is the primary symptom in many cases, prompting a thorough investigation to find the disorder's cause.[8] SLCTs are mainly ovarian upon diagnosis, usually unilateral solid tumors (only 1.5% occur bilaterally).

The histologically distinctive reinke crystals are present in SLCTs, which are classed as well-differentiated, intermediately differentiated, and undifferentiated based on the proliferation of Sertoli and Leydig cells in varying proportions.[10]
They appear as encapsulated solid enhancing masses on MRI and CT scans, and they are echogenic solid masses in ultrasound imaging.[11]

It is classified as granulosa cell tumour, SLCT group/androblastoma, gynandroblastoma, unclassified group. SLCT group has been divided into Sertoli cell tumour, SLCT group, Leydig cell tumour group and Hilus cell tumour. In our first case, presented with postmenopausal virilisation features, with elevated serum total testosterone levels and no significant tumour in radiological imaging was detected.

Second case with postmenopausal bleeding and on evaluation, found a large right solid ovarian mass on imaging. Proceeded to laparoscopic hysterectomy with bilateral salpingoophorectomy and histopathological report came as sex cord stromal tumour, first one as Leydig

cell tumour group and second one as Sertoli-Leydig cell group with immunohistochemistry positive.

CONCLUSION

SLCT poses a diagnostic challenge dueto its rarity. Early stage SLCTs have a favourable prognosis. An accurate diagnosis based on clinical, radiological and pathological features have important therapeutic implications. In young patients who wants to preserve fertility, can go for bilateral salpingoophorectomy. Hysterctomy with bilateral salpingoophorectomy is the treatment of choice in post-menopausal patients. The overall 5-year survival rate was reported as 100% for well-differentiated and a collective 80% for moderately and poorly differentiated.

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