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Case Report

A rare ovarian Leydig cell tumour masquerading as postmenopausal bleeding: a case report

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ABSTRACT

Ovarian Sertoli-Leydig cell tumors (SLCTs) are rare ones, accounting for less than 0.2% of ovarian malignancies. While typically associated with virilization, they can present atypically with postmenopausal bleeding. We report a 72-year-old postmenopausal woman with post-menopausal bleeding. The diagnosis was endometrial hyperplasia. The patient, with a history of diabetes, hypertension, and dyslipidemia, had postmenopausal bleeding lasting five days without features of virilization. Ultrasonography showed a bulky uterus with a fibroid, adenomyoma, and thickened endometrium. Endometrial sampling revealed focal hyperplasia without atypia. Despite progestin therapy, she had recurrent vaginal bleeding. Hence decision for surgical management was sought. Histopathology confirmed a 1 cm Leydig cell tumour in the right ovary, with no significant findings in the left ovary or tubes.

Keywords: Sertoli-Leydig cell tumors, Postmenopausal bleeding, Endometrial hyperplasia, Virilization

INTRODUCTION

Ovarian Sertoli Leydig cell tumors (SLCT) are rare sex cord-stromal tumors. They account for <0.2 % of all ovarian malignancies. These tumors are mainly seen in young females at around 23 years of age, but can occur in any age group.¹ Clinical presentations range from asymptomatic to, with features of extreme virilization. About 50% of patients present with features of virilization.² Signs of virilization can be amenorrhoea, hirsutism, male pattern of alopecia which may be either due to testosterone secreted from the tumour or by peripheral conversion of estrogen.²

It is difficult to diagnose SLCT radiologically, and diagnosis is confirmed only by histopathology. Mostly these tumors occur unilaterally, and the treatment is surgical. Fertility sparing surgeries are considered in fertile women and total hysterectomy with bilateral salpingo oophorectomy may be considered in older patients.² Below we present an atypical presentation of SLCT, in a 72-year-

old post-menopausal lady who came with postmenopausal bleeding and endometrial hyperplasia, with no features of virilization. Diagnosis was confirmed only after histopathology report.

CASE REPORT

A 72-year-old postmenopausal lady, a known case of type 2 diabetes mellitus, systemic hypertension and dyslipidemia presented with postmenopausal bleeding one month back. Bleeding lasted only for 5 days. It was not associated with pain and there was no history of heavy bleeding. She had history of similar bleeding one year back but not evaluated.

On examination she was moderately built and nourished. Had no features of virilization. Her general examination and vitals were within normal limits. No mass on abdominal examination. Gynaecological examination showed a healthy, but atrophic cervix and a normal sized

mobile uterus with fornices free. No adnexal mass detected.

Blood investigations were within normal limits. Ultrasound report showed bulky uterus with anterior wall fibroid and an adenomyoma 4×3.2 cm. also a borderline thickened endometrium with thickness 5.5 mm. Dilatation and curettage were done and histopathology reported as disordered proliferative endometrium with focal hyperplasia without atypia. She was managed with medroxy progesterone acetate for one month and had bleeding on withdrawal. She was planned for and underwent total abdominal hysterectomy with bilateral salpingo oophorectomy. Uterus found regularly enlarged to 10 weeks gravid uterus size. Bilateral fimbriae not visualized. Post sterilization status adhesion of fimbrial end to omentum. Bilateral ovaries were atrophic.

Histopathology showed chronic cervicitis and adenomyosis. Gross examination of right ovary showed a tumour of size measuring 1 cm (Figure 1). Histopathology examination showed individual neoplastic cells in oval to polygonal shape with abundant eosinophilic to clear cytoplasm (Figure 2). Low power showed neoplastic cells arranged in sheets, lobules, and nests (Figure 3). Left ovary and both tubes showed no significant pathology.



Figure 1: Gross examination of right ovary showing the tumour.

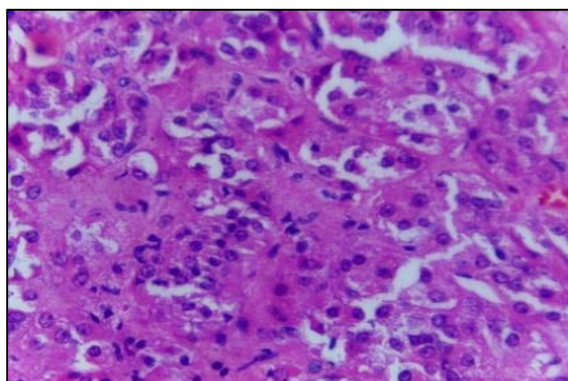


Figure 2: Individual neoplastic cells in oval to polygonal shape with abundant eosinophilic to clear cytoplasm.

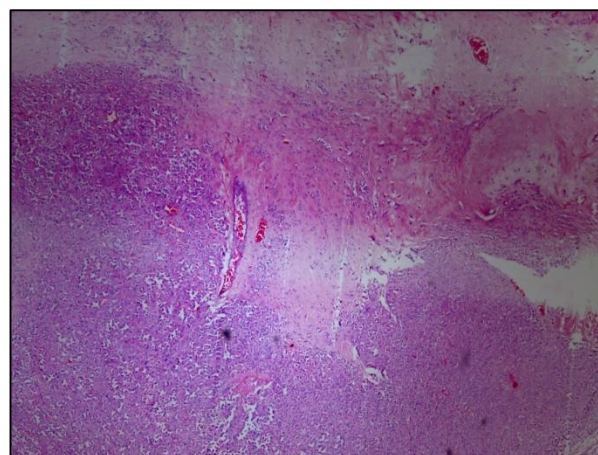


Figure 3: Neoplastic cells arranged in sheets, lobules, and nests.

DISCUSSION

Sertoli-Leydig cell tumors are extremely rare neoplasms. They account for only about 1% of sex cord stromal tumors and less than 0.2% of all primary ovarian tumors. They are unilateral in 98% of the cases.³ Sertoli Leydig cell tumors most commonly present around 25 years of age. But 10% of the case present before menarche and after menopause.⁴ the most common presentation is that of symptoms of virilization which is seen in around 77% of the cases.⁵ post-menopausal bleeding, endometrial hyperplasia, carcinoma endometrium are among the rare presentations. It may be either due to the estrogen secreted by the Sertoli cells or by the peripheral aromatase cytochrome p450 mediated conversion of testosterone into estrogen.⁴

Hyper androgenism (raised serum testosterone more than 3times the upper limit) in the backdrop of a normal serum DHEA-S (which excludes adrenal hyperandrogenism), leaves ovarian androgen secreting tumor as the probable diagnosis.⁶ But absence of hyperandrogenism or virilization cannot exclude Leydig cell tumour as in 25-30% cases serum androgen levels are normal.⁷

Imaging with USG, CT, MRI are useful in diagnosis. Ultrasound is the best imaging for SLCTs and colour doppler is used to characterise SLCTs as they are usually highly vascular tumors. CT can show well defined tumors and T2 MRI can show low signal intensity due to fibrous stroma.⁸ About 20% SCLTs are missed on imaging due to very small size.³ PET scan with fluorodeoxyglucose as marker can detect small focus of Leydig cell tumor.⁹

Sertoli-Leydig cell tumors are classified into-well differentiated, moderately differentiated, poorly differentiated, with a retiform pattern, with heterologous elements (mucoprotein, focal carcinoid elements, cartilage, etc), and mixed. Generally, all the well differentiated tumors are benign, and tumors other than well differentiated and tumors with retiform or

heterologous elements pattern can behave in a malignant manner.⁸ 20% of Sertoli-Leydig cell tumors has heterologous elements. They present in some poorly or intermediately differentiated tumors, predominantly seen as cystic tumor.⁸ The gold standard treatment for SLCTs is surgery. For woman with a completed family or does not want to preserve fertility, total hysterectomy, and bilateral salpingo-oophorectomy or cytoreductive operation should be recommended. A unilateral salpingo-oophorectomy may be done in the early stages for young patients as a fertility preservation procedure.¹⁰

CONCLUSION

SLCTs, although typically seen in younger women, can occur in postmenopausal period. Endometrial hyperplasia may result from estrogen secretion or peripheral conversion of androgens. This case highlights the importance of considering SLCT in atypical presentations of postmenopausal bleeding and supports surgical management as the treatment of choice.

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Ethical approval: Not required

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