

## Bilateral Leydig Cell Tumor of Ovary: A Rare Case Report

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### Abstract

*Background:* Leydig cell tumors of the ovary a rare benign neoplasms, usually occur in postmenopausal women associated with symptoms of virilization. Only five cases of benign Leydig cell tumor have been reported in literature [1].

*Case Report:* Here is a case of Leydig cell tumor of ovary. A 58 yr old female referred from Endocrinology department due to a biochemical diagnosis of hyperandrogenism. Serum Testosterone levels were 5.36 ng/ml. The patient presented with alopecia and hirsutism since 12 yrs. Ultrasonography showed right ovary measuring 2.8x1.8 cm with isoechoic lesion, left ovary measuring 3.8x2.6 cm. CT scan showed subserosal fibroid measuring 3x3.6 cm in fundus of uterus both ovaries were in postmenopausal status. Hysterectomy with bilateral salpingo-oophorectomy was performed.

*Results:* Histopathological finding revealed bilateral Leydig cell tumor, measuring 2.0x1.5 cm in the right and left ovary. After surgery serum testosterone level were in normal limits and there was regression of signs of virilization.

*Conclusion:* Bilateral Leydig cell tumor are rare, usually benign, they require surgical treatment and followup.

**Keywords:** Ovarian tumors; Virilization; Bilateral Leydig cell tumor.

### Introduction

Leydig cell tumor is a rare sex cord stromal group accounts for less than 0.1% of all ovarian tumors. They are usually benign, unilateral and characterized by the presence of crystals of Reinke in the steroid cells. They are functional tumors and one of the ovarian virilizing tumors. The average age of occurrence is 58 years and are commonly hormone producing mainly androgenic and rarely oestrogenic. They produce testosterone, leading to hyperandrogenism and virilization such as hirsutism, voice deepening, clitoromegaly, increased muscle mass in postmenopausal women [2]. Very rare case report of a bilateral Leydig cell tumor as a cause of virilisation in postmenopausal women is being presented. Hysterectomy with bilateral salpingo-oophorectomy was performed.

### Case Report

A 65 yr old female, complaints of excessive hair growth on face mainly chin, upper lip, chest and on abdomen, and there is also history loss of hair over scalp since 12 yrs. There is no significant past and family history. Obstetric history-Married at the age of 20 yrs, did not conceive for 9 yrs after

marriage. P4L3D1 with normal menstrual cycle. Attained menopause 19 yrs back. There is no history of hirsutism in her reproductive age group. On physical examination, signs of virilization, such as beard which required shaving twice a day and excessive hair growth also seen on chest and abdomen (28 on Ferriman gallway scale), alopecia and enlarged clitoris. Ultrasonography revealed mild thickened endometrium-7 mm with few cystic areas and right ovary measuring 2.8x1.8 cm with isoechoic lesion, left ovary measuring 3.8x2.6 cm. CT scan showed subserosal fibroid measuring 3x3.6 cm in fundus of uterus both ovaries were in postmenopausal status. Endocrinological workup was done: (Table 1) Hysterectomy with bilateral salpingo-oophorectomy was performed and specimen submit for histopathology examination.

Table 1:

Harmones		Reference
Testosterone	5.36 ng/ml	0.06-0.82 ng/ml
Free testosterone	4.69 pg/ml	0.001-7.01 pg/ml
Dihydrotestosterone	430 pg/ml	0.13-7.01 pg/ml
17 alpha hydrotestosterone	1.40 ng/ml	0.13-0.51 ng/ml
Adrenocorticotropic	56.6 pg/ml	10-50pg/ml
Dehydroepiandrosterone	4.7 ng/ml	2-6 ng/ml

Grossly circumscribed greyish brown tumor on cut section measuring 2.0x1.5 cm in both right and left ovaries.

*Microscopically:* Tumor cells are arranged in lobules separated by fibrous septa. The tumor cells are polygonal with abundant eosinophilic cytoplasm and round nuclei with prominent nucleoli. Clustering of nuclei, creating intervening eosinophilic nuclear free zone. Also seen rod like elongated eosinophilic inclusion (Reinke crystals) in cytoplasm and the diagnosis of Leydig cell tumor made.



Fig. 3: Clinical photograph showing clitoromegaly



Fig. 4: USG Showing Bilateral well defined hypoechoic solid adnexal lesions with few cystic areas

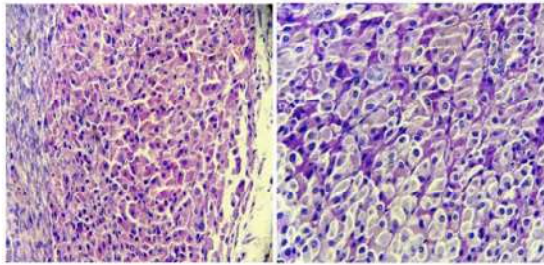


Fig. 1 & 2: Clinical photograph showing before and after signs of virilisation

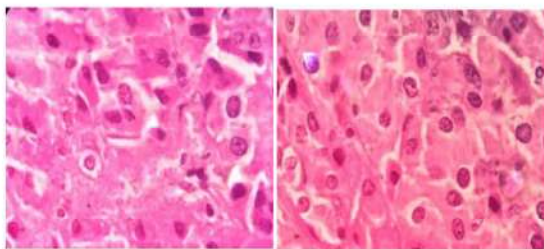


Fig. 5: Grossly circumscribed greyish brown tumor on cut section measuring 2.0x1.5 cm in both right and left ovaries





**Fig. 6 (10x) & Fig. 7 (40x):** Photomicrograph showing tumor cells are polygonal with abundant eosinophilic cytoplasm with prominent nucleoli. Clustering of nuclei, creating intervening eosinophilic nuclear free zone



**Fig. 8 & Fig. 9:** Photomicrograph showing tumor cells with Reinke's crystals

### Discussion

Hirsutism occurs mainly due to excessive androgen secretion and increased peripheral conversion of weaker androgen to more potent molecules [3]. Signs of virilization involves severe hirsutism, frontal balding, deepening of voice, increased muscle mass, breast atrophy, altered body fat and clitoromegaly [4]. The serum testosterone levels above 3.5 nmol/l in postmenopausal women, the origin is usually an ovarian tumour or hilar cell hyperplasia [3]. In the present case the main clinical data features and reason for consultation were hyperandrogenic symptoms. Ovarian tumors associated with hyperandrogenism are primitive tumors of the sexual cords and stroma (granulosa cell tumor, thecoma, sertoli Leydig tumors), steroid cell tumors includes luteomas, Leydig cell tumors and others are primary and secondary tumors containing functioning stroma [5]. Leydig cell tumors are rare ovarian steroid cell neoplasm composed mainly of Leydig cells. They are of two types hilar type and non hilar type. More than 75% of these patient shows signs of virilization by increase production hormone mainly testosterone and rarely present manifestation of hyperestrogenism including endometrial carcinoma and occasionally these tumors can be inactive [6]. In the present

case the diagnosis is mainly based on signs of virilization, increase in the serum testosterone levels. Hysterectomy with bilateral salpingo-oophorectomy was performed. Grossly tumors centred in hilar region, microscopically, tumor cells have abundant eosinophilic granular cytoplasm with lipochrome pigment and the presence of Reinke's crystals is characteristic but not pathognomonic. The postoperative followup done, serum testosterone level decreased after 3 wks. The diagnosis of hormone producing ovarian tumors is based on clinical, biological and imaging techniques. However the imaging techniques are insufficient because of smaller tumor size as in present case. Therefore if any ovarian tumor is suspected clinically and biologically surgery is indicated.

### Conclusion

In conclusion, bilateral Leydig cell tumors of ovary are rare, usually benign; they require surgical treatment. However in spite of its low incidence bilateral Leydig cell tumor should be considered in a patient in postmenopausal age group with signs of virilization.

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