Case Report

A Young Female with Secondary Amenorrhea and Hirsutism: A Rare Case Report of Steroid Cell Tumor of the Ovary

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ABSTRACT

Objective: To report a case of a young female with longstanding secondary amenorrhea and hirsutism, ultimately diagnosed with an ovarian steroid cell tumor.

Methodology: A comprehensive review of the patient's clinical presentation, biochemical profile, radiological findings, surgical intervention, and histopathology was undertaken.

Results: A 25-year-old female presented with secondary amenorrhea and progressive hirsutism for five years, accompanied by weight gain and elevated blood pressure for two years. She reported regular menstrual cycles until 2015. Clinical examination revealed a Ferriman-Gallwey score of 25, without signs of proximal myopathy or striae. Laboratory investigations showed markedly elevated serum testosterone (527 ng/dL), low luteinizing hormone (LH 0.14 mIU/mL), and low follicle-stimulating hormone (FSH 0.76 mIU/mL). Thyroid function tests and DHEA-S levels were within normal limits. Serum 17-hydroxyprogesterone (17-OHP) was elevated (10.9 ng/mL), and the overnight dexamethasone suppression test (ODST) was normal (0.70 µg/dL). Pelvic CT imaging revealed a left ovarian mass adherent to the sigmoid colon and in close proximity to the uterus and contralateral ovary. She underwent exploratory laparotomy, left salpingo-oophorectomy, and total omentectomy for tumor removal. Postoperatively, serum testosterone and 17-OHP levels normalized, and the patient resumed menstruation.

Conclusion: In women presenting with signs of virilization, a thorough evaluation is essential to differentiate between adrenal and ovarian sources of androgen excess. Although rare, ovarian steroid cell tumors should be considered in the differential diagnosis of hyperandrogenism in adult females.

KEY WORDS: Steroid cell tumor, Secondary amenorrhea, Hirsutism, Virilization, Hyperandrogenism.

INTRODUCTION

Scully described the term "steroid cell tumor of the ovary" in 1979.[1] These tumors are divided into three subtypes according to their cells of origin: stromal luteoma, leydig cell tumor and steroid cell tumor, not otherwise specified (NOS). Out of these, the steroid cell tumors, NOS account for about 56% of steroid cell tumors. The incidence of steroid cell tumors, NOS is more common in women of child-bearing age group, especially the third and fourth decades.[2] A majority of steroid cell tumor NOS are unilateral and well circumscribed and their size varies from 1.2 to 45 cm.[2] NOS, sometimes produce hormones other than testosterone, for example, estradiol secretion has been reported in 6–23% of patients. These tumors have been associated with Cushing's syndrome in 6–10%

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of cases.[1,3-4] Preoperative evaluation for women with virilization includes a bimanual examination, serum testosterone, DHEA-S, and computerized tomography (CT) of the adrenals and ovaries.[5] Ultimate diagnosis is made by histology. Microscopically, the neoplastic cells have abundant clear vacuolated cytoplasm with rich vascularity, and absence of atypia or necrosis. As far as immunohistochemical markers are concerned, inhibin is quite useful in differentiating this tumor from other non sex cord tumors.[6,7]

CASE REPORT

We report the case of a 25 years old female patient who presented with secondary amenorrhea, hirsutism for 5 years, raised blood pressure for 2 years and weight gain.

On examination, Her blood pressure was 149/103 mmHg, pulse 72 beats/min. BMI was 27.6 (height: 168 cm, weight: 68 kg).There was no straie, proximal myopathy, acanthosis nigricans, The thyroid gland was not palpable, and no lymphadenopathy was noted. Systemic examination was completely unremarkable. Hirsutism was present on face and body with Ferriman Gallwey score of 25. Mild temporal baldness was present.

CT pelvis with contrast was done which revealed left ovarian mass adherent to the sigmoid colon abutting the fundus of uterus and right ovary, no evidence of peritoneal disease, abdominopelvic lymphadenopathy or distant metastasis noted. She underwent laparotomy,left salpingo-oophorectomy, excision of left para tubal cyst, infracolic omenectomy and bilateral pelvic lymphadenectomy. Histopathology of left ovarian mass turned out to be consistent with steroid cell tumor with following features.

Gross features: The ovarian mass had smooth outer surface and the capsule was intact. Cut surface was lobulated, firm, yellow with few brown areas.

Microscopic features: The ovarian mass showed a neoplastic lesion arranged in diffuse sheets of polygonal cells. The cells had abundant amount of granular, lightly eosinophilic to clear cytoplasm with intracytoplasmic vacuoles. The cells have distinct cell borders. The nuclei were small, round and uniform and with inconspicuous nucleoli. No significant nuclear atypia, increased mitoses or any other features of malignancy were seen. The morphological features favored the diagnosis of "Steroid Cell Tumor".

Postoperative follow-up: The thyroid gland was not palpable, and no lymphadenopathy was noted, it was 25.51 ng/dl (normal is 8.4 -48.1 ng/dl). 3 months after

Table-I: Laboratory Investigations.

Investigations	*Patient's value*	*Normal values*
Hemoglobin Hematocrit R.B.C.	18.2 53.7 6.46	(11-14.5) (34-45) (3.61-5.2)
Erythropoietin level	2.44	(4.3-29 is normal)
JAK 2 mutation	Negative Due to polycythemia, hematology opinion was taken. Phlebotomy of 1 pint was done , after which Hb reduced to 15.2	
Prolactin	23.3 ng/dl	3.8-23 ng/dl
LH	0.14 mIU/ml	1.7-15 mIU/ml
FSH	0.76 mIU/ml	1.4-9.9 mIU/ml
Total Testosterone	526.9 ng/dl	8.4-48 ng/dl
TSH	2.8 uIU/ml	0.4-4.0 uIU/ml
FT4	0.98	0.9-1.87
DHEAS	147 ug/dl	35-430ug/dl
17-OH Progesterone	10.9 ng/ml	0.15 - 0.70 ng/ml
HBA1c	6.1	< 5.8 is normal
Cortisol (ODST)	0.70	<1.0

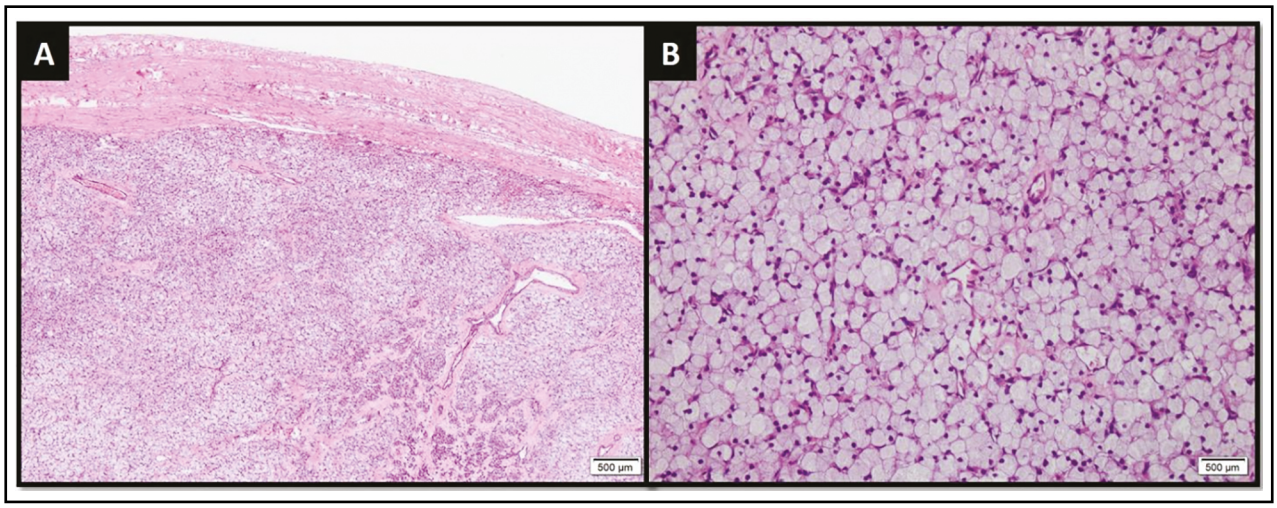

Fig.1: [A] Low power view of tumor showing cells arranged in sheets. [B] The neoplastic cells are polygonal with distinct cell borders and abundant clear cytoplasm. The nuclei are small, round and uniform.

surgery her 17-OH Progesterone level was checked, turned out to be 0.50 ng/ml (normal is 0.15 -0.70 ng/ml) with repeat Testosterone level of 46.36 ng/dl. She returned for a follow-up visit at the endocrine clinic. Her menstrual cycles had resumed, and hirsutism had resolved.

DISCUSSION

Steroid cell tumor, not otherwise specified (NOS) can present as abdominal pain but the more significant clinical manifestations of this type of tumor are associated with the hormonal activity and virilizing properties, occurring in 56-77% of patients.[1] It may cause precocious puberty in children, and in adults it can manifest as oligomenorrhea, hirsutism, acne, increased libido, and deepening of the voice.[1,3,4] Usually these tumors are benign, but 25-43% are clinically malignant, with 20% of cases found to exhibit metastasis outside of the ovary usually within peritoneal cavity and rarely occur at distant sites.[3,4,8] A study by Hayes and Scully reported five pathological features predictive of malignancy: two or more mitosis per 10 high-power fields, necrosis, size of the tumor (more than 7 cm), hemorrhage, and grade 2 or 3 nuclear atypia.[1] Androgenic manifestations are common in these tumors as they secrete hormones like androstenedione, α-17 Hydroxy Progesterone, and testosterone.[9,10] The treatment of these tumors should be based on the histological picture, surgical staging and patient's desire to preserve fertility.[1,12] In young patient who want to preserve their fertility, unilateral salpingo-oophorectomy is the preferred method of treatment.[12] For women who have completed childbearing, total abdominal hysterectomy with bilateral salpingo-oophorectomy and complete surgical staging is indicated.[3,8] The persistently raised 17 Hydroxy Progesterone levels in our case was due to steroid cell tumor of ovary after excision of which the levels get normalized .

Regular follow-up evaluation with measurement of serum testosterone level is mandatory, but since little is known about the behavior of these tumors, it is not determined for how long.[8,4]

CONCLUSION

In women presenting with signs of virilization, a thorough evaluation is essential to differentiate between adrenal and ovarian sources of androgen excess. Although rare, ovarian steroid cell tumors should be considered in the differential diagnosis of hyperandrogenism in adult females.

Conflicts of Interests: The authors declare no conflict of interest.

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Author's Contributions:

All authors certify that they have participated sufficiently in the work. All participated in the conception of the work, with the analysis and interpretation of data. Sabiha Banu composed the manuscript, that was revised by Owais Rashid and Abdul Aziz. All authors approved the final version of the manuscript and agree to be accountable for all the aspects of the work.

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