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Case Report Open Access

A Rare Case of Ovarian Sertoli-Leydig Cell Tumor in Postmenopausal Woman Presenting with Alopecia

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Abstract

Mild hirsutism and alopecia in postmenopausal women can be a normal physiological response. Sex cord stromal tumors of ovary account for approximately 5-8% of all ovarian tumors. When hirsutism is accompanied by signs of virilization such as severe balding, deepening of voice or clitoromegaly, an underlying androgen-secreting tumor, that may be malignant must be ruled out. We report a rare case of 46 year old female with premature menopause and symptom of hair loss. She had high testosterone levels and left ovary mass. As Cushing syndromes and late onset congenital adrenal hyperplasia were ruled out, an ovarian source of androgen was suspected. She underwent hysterectomy with bilateral salpingo-oophorectomy. A diagnosis of left ovarian sex-cord stromal tumor favoring Sertoli-Leydig cell tumor was confirmed.

We report this case for its rare nature and atypical presentation.

Keywords: Hirsutism; Virilization; Sertoli-Leydig cell tumor

Introduction

Mild hirsutism and alopecia in postmenopausal women can be a normal physiological response: with reduction of ovarian follicles, there is diminished secretion of estrogen and progesterone, increasing the impact of androgen on sebaceous glands and hair follicles [1]. The post-menopausal ovary remains hormonally active, secreting sufficient amounts of androgens and estrogens, many years after menopause [2]. Post-menopausal virilization may result from adrenal tumors, including androgen-secreting carcinomas and adenomas; from ovarian tumors, including Sertoli-Leydig cell tumors, granulosa-theca cell tumors and hiluis cell tumors; or from benign ovarian conditions such as ovarian stromal hyperplasia and hyperthecosis [3]. When hirsutism is accompanied by signs of virilization such as severe balding, deepening of voice or clitoromegaly, an underlying androgen-secreting tumor that may be malignant must be ruled out [4].

We are reporting here a rare case of virilizing androgenic tumor of ovary in a postmenopausal woman with only complaint of severe alopecia.

Case Presentation

A 46 year old female presented in our Endocrinology clinic with presenting complaint of severe hair-loss. This was her sole complaint. She gave history of attaining menopause at a premature age of 39 years. There was no relevant past surgical history. Her obstetric history was insignificant with G3P3A0 and all were full term normal deliveries. She was not on any medication at the time of her presentation. Her family history was significant with mother hypertensive and father diabetic. Social history for smoking and drinking was negative.

On physical examination, BMI was 28.16 kg/m^2, her pulse was 80 beats per minute, regular, blood pressure recorded was 120/80 mmHg. She had acanthosis and skin tags. Temporal hair recession was prominent. Except for male- pattern of alopecia there was no other sign of virilization. Her Ferriman-Gallaway score was 4. Her face was neither Cushingoid nor acromegalic.

Her laboratory work up revealed following results: Serum total testosterone 6.5ng/ml (Normal range for females: 0.3-0.9 ng/ml) done by Radio Immuno Assay method, Serum DHEAS 4300 ng/ml (Normal range for females: 1000-2600 ng/ml) by RIA method, serum Thyroid Stimulating Hormone 2.8 MIU/ml, Serum ferritin 49.3 ng/ml, fasting blood glucose 82.1 mg/dl , fasting insulin levels were 26m IU/ml. Her lipid profile was as follows: serum cholesterol 181.2 mg/dl, High Density Lipoprotein cholesterol 39.5 mg/dl, Low Density Lipoprotein cholesterol Direct 87.0 mg/dl, serum triglycerides 142.6 mg/dl. And Very Low Density Lipoprotein 28.5 mg/dl. Cushing Syndrome was ruled out as her 8.a.m. serum cortisol levels were 8 µg/dl (Normal range 5-25 µg/dl). Overnight dexona suppression was done to rule out Cushing Syndrome (Serum Cortisol overnight dexona suppression was 3 microgram/dl) and normal 17-hydroxyprogesterone (to rule out late onset congenital adrenal hyperplasia).

Since her testosterone levels were very high, the test was repeated which showed same result.

As the patient was found out to be insulin resistant, she was put on Metformin 1000 mg/day and was advised diet and lifestyle modification. She was advised complete gynecological checkup. Her gynecological examination revealed fullness in right fornix and minimal cervical erosion. Ultrasound showed uterine dimensions: 11 × 6.8 × 5.2 cms. Endometrium 4.1 mm thick and hyper-echoeic in nature. Myometrium was non-homogeneous with signs of adenomyosis. There was no evidence of any space occupying lesion in uterus or fibroid. Left ovary showed chocolate cyst.

She underwent hysterectomy with bilateral salpingo-oophorectomy. Histopathology report turned out to be left ovarian tumor, benign in nature. Cells from the tumor showed columnar cells arranged in pseudorossets, round polygonal cells in sheets. Cells had moderate amount of granular and at places clear cytoplasm. Nuclei showed moderate anisonucleosis and powdery chromatin. Findings revealed left ovarian sex-cord stromal tumor favouring Sertoli-Leydig cell tumor.

Discussion

Sex cord stromal tumors of ovary account for approximately 5-8% of all ovarian tumors. They are composed of granulosa cells, theca cells, sertoli cells, leydig cells and fibroblasts singly or in variable combinations [5,6]. Leydig cell tumor is a rare form and accounts for less than 0.1% of all ovarian tumors [7] and are usually benign and unilateral [8,9].

In many of the cases reported, the age of the patients is more than 60 [10,4,11]. Our patient is much younger as compared to them. However same age onset is reported by 2 other studies [6,12]. Our case was rarest of rare as the presenting complaint was excessive hair loss alone. There was no other symptom suggestive of virilization or hyperandrogenism. Very high levels of testosterone and normal levels of cortisol and 17 Hydroxyprogesterone (17-OHP) ruled out Cushing Syndromes and late onset CAH as cause of alopecia. Transvaginal ultrasound revealed chocolate cyst of left ovary and diagnosis confirmed after hysterectomy with bilateral salpingo-oophorectomy on histopathology.

Diagnosis of hyperandrogenism in postmenopausal women is challenging. Postmenopausal virilization may be associated with adrenal or ovarian androgen-secreting tumors or benign hyperplasia. Imaging techniques do not always reveal the cause of hyperandrogenism and may be misleading [4]. A detail clinical history and lab investigations can be instrumental in clinching proper diagnosis as in our case.

Estrogen levels drop abruptly after menopause whereas androgen secretion declines during the reproductive years. Subsequently an imbalance among estrogens and androgens during menopause, amplified by a decrease in sex-hormone binding globulin (SHBG) concentrations may result in hyperandrogenic symptoms [13].

Androgen secretion in pre- and post-menopausal ovaries depends on LH stimulation. The very high gonadotrophin levels of menopause could maintain ovarian androgen production as a result of which, menopause may be accompanied by decrease in body and scalp hair and can be considered as a part of normal menopausal process. The development of true hirsutism, alopecia or acne should not be considered normal in post-menopausal women. When signs of virilization such as severe balding, deepening of voice or clitoromegaly is present, an underlying androgen secreting tumor must be ruled out [14]. We report this case for its rare nature and atypical presentation.

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