

A Rare presentation of androgen secreting adrenal adenoma with malignant ovarian tumour

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Abstract:

Adrenal and ovary are the source of androgens in female. We submit a case report of a fifty two year old, nulliparous, post menopausal woman, who presented with clinical features of virilization and abdominal distension. On further evaluation, she was found to have bilateral complex ovarian mass with adrenal mass. Blood investigation showed elevated androgens with normal adrenal hormones. The differential diagnosis of synchronous ovarian tumour with functioning adrenal carcinoma/adenoma causing virilisation or malignant ovarian sex cord-stromal tumours with adrenal metastasis was made.

Patient underwent staging laparotomy with total abdominal hysterectomy with bilateral salpingo-oophorectomy and left adrenalectomy.

HPE showed bilateral endometrioid adenocarcinoma of ovary with metastasis to peritoneum and omentum with adrenocortical adenoma. Adrenal adenoma is the source of androgen. It was dual pathology which is uncommon and Adrenal adenoma that secretes androgens exclusively are extremely rare.

Key-words: virilisation, adrenal adenoma, adrenalectomy

Key Messages : Its a case of bilateral endometrioid adenocarcinoma of ovary with metastasis to peritoneum and omentum with adrenocortical adenoma. Adrenal adenoma is the source of androgen. It was dual pathology which is uncommon and Adrenal adenoma that secretes androgens exclusively are extremely rare.

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I. Introduction:

Both Adrenal and ovarian tumours can cause virilization. We are presenting a case where there is both adrenal and ovarian tumours with virilisation. It is difficult to differentiate whether androgen is from adrenal or ovary. Synchronous ovarian tumour with adrenal tumour is very rare. Adrenal adenoma that secretes androgens exclusively are extremely rare. This is a first case to report a dual pathology of androgen secreting adrenal adenoma with ovarian malignancy.

II. Case History:

CASE REPORT:

Fifty two year old, postmenopausal, nulliparous woman was admitted in our hospital with abdominal distension since two months increased male pattern of hair growth for past one year. Patient had deepening of voice. Patient is a known hypertensive since 5 yrs and controlled with medication.

On examination she was afebrile, PR-75, BP-120/80 mmHg. Increased hair growth seen over face, lip, upper arm, inner thigh, upper & lower back and androgenic alopecia was present (figure 1a). She was anemic. Per abdominal examination showed distension of abdomen, there was a mass measuring 18x12 cm involving suprapubic right and left iliac fossa and umbilical region. The mass was firm to hard in consistency with irregular surface. All borders are well made out except the lower border. External genitalia examination showed normal external meatus and clitoromegaly (figure 1b). Per vaginal examination revealed normal cervix with mass felt mostly over the right and the posterior fornix.

Investigations showed normal CBC, RFT, LFT.

HORMONAL ASSAY: CA125-4333 u/ml (<35 u/ml) T. TESTOSTERONE-171.76 mg/dl (14-76 mg/dl) F. TESTOSTERONE -86.4 pg/ml (0.9 pg/ml) DHEAS - >1500 mg/ml (48-361 mg/ml) 17 OH Progesterone-2.3 ng/ml

24 hrs cortisol-30 mcg (10-100 mcg/24 hrs), Urinary metanephrine-50 mcg (30-180 mcg/24 hrs), urinary nor metanephrine-150 mcg (128-485 mcg/24 hrs)

USG abdomen -showed right kidney was measuring 10x4.4cm and normal CMD and PCS. Left kidney was measuring 10.2x3.8cm and normal CMD and PCS. 5x 7 cm mixed echoic lesion seen over the superior pole of left kidney. 20x15 cm mixed echoic mass extending from pelvis to epigastric region with cystic and solid areas. Both ovaries could not be visualised separately. Other intra abdominal organs were normal

CECT abdomen- heterodence mass measuring 15x8cm arising from right side of pelvis and right ovary could not be seen separately. There is other heterodence mass measuring 10x7cm arising from left side of the pelvis and left ovary could not be seen separately. Bilateral kidneys normal. There is a heterodence mass measuring 7x4 cm arising from left adrenal. Other intra abdominal organs were normal. (figure 1c&1d)

The differential diagnosis including synchronous ovarian tumour with functioning adrenal carcinoma/adenoma causing virilisation or androgen secreting malignant ovarian sex cord-stromal tumors with adrenal metastasis. Patient underwent staging laparotomy with total abdominal hysterectomy with bilateral salpingo-oophorectomy and left adrenalectomy (figure 2a&2b). Adrenalectomy is done because adrenal mass could be a androgen secreting carcinoma/adenoma or a solitary metastasis from ovarian tumour and the adrenal mass measures around 7 cm which itself is a indication for intervention

HPE showed bilateralendometrioid adenocarcinoma of ovary with metastasis to peritoneum and omentum with adrenocortical adenoma. IHC shows synatophysin-negative, s 100-negative, cytokeratin-positive, ki 67-less than one percentage. Adrenal adenoma is the source of androgen. (figure 2c&2d)

III. Discussion

Virilizing adrenal tumours are a rare entity. Their clinical manifestations vary depending on age at onset. The most common adrenal mass that hypersecretes sex steroid is an adrenal carcinoma that concomitantly exhibits cortisol hypersecretion. Adrenal tumors that secrete androgens exclusively are extremely rare. Approximately 50% of such lesions ultimately proven to be benign. Ovarian sex cord-stromal tumors can produce androgen as well. In this case DHEAS is more than 1500 mg/ml (48-361mg/ml) and is very high. DHEAS usually of adrenal origin and very rarely secreted by ovary. Patients who presented with virilization, dexamethasone suppression testing was used to differentiate adrenal androgen excess from an ovarian androgen source. This approach has proven unreliable and has been largely replaced by radiographic imaging modalities. In patients with suspected adrenal and ovarian tumors, selective venous catheterization of the adrenal and the ovarian veins for selective measurement of androgens is helpful in case with uncertain localization. Surgery is the only therapeutic option with curative intention for patients with sex hormone-producing tumors. Only complete removal of the tumor tissue offers definitive cure.

IV. Conclusion

synchronous ovarian tumour with adrenal tumour very rare. Adrenal adenoma that secrete androgens exclusively are extremely rare. This is a case of adrenal adenoma and B/L endometrioid adenocarcinoma of ovary with metastasis to peritoneum and omentum. This is a first case to report a dual pathology of androgen secreting adrenal adenoma with ovarian malignancy.

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