

Testosterone and cortisol co-secretion by an adrenocortical adenoma presenting as secondary polycythemia



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Background

Androgen co-secretion in patients with adrenal Cushing's syndrome is considered a marker of malignancy.

Mixed secretion by benign adrenocortical tumors is rare (1-2).

We describe a case of a middle aged woman presenting with Cushing's syndrome and polycythemia.

Case report

A 57-year-old lady was referred by the Hematology Service to our Endocrine Clinic for evaluation of an adrenal mass found in an abdominal CT in the course of investigating secondary polycythemia.

The patient, a 35 pack-year-smoker, had presented with upper arms petechiae and a hematocrit of 52 and hemoglobin of 17, while it was up to 42 up to 5 years previously.

Jak2V617F mutation analysis was negative. Her past history was notable for natural menopause at age 45, recent onset hypertension and dyslipidemia. She had noted worsening hirsutism on her trunk and arms.

On clinical examination the patient had the classic Cushingoid features, Ferriman Gallwey score 17, marked female pattern hair loss but no other signs of virilization.

The pertinent lab findings were as follows:

Hb	16.3 g/dl
Ht	49.1%
K	4.8
Glu	115 mg/dl
ALT	29 mg/dl
Cortisol after overnight dexamethasone suppression	483nmol/l (<50nmol/l)
24h UFC	405
ACTH	1 pg/ml (7.2-63.6)
Aldosterone	6.24ng/dl (4-31)
Plasma renin activity	0,17 ng/ml/h (0.5-4.7)
Estradiol	18 pg/ml (<50)
Testosterone	3.49nmol/l (0.43-1.24)
Δ 4-androstenedione	3.4 ng/ml (0.3-3.3)
DHEAS	50.8
17OH-progesterone	2.64 ng/ml (0.2-1)
Erythropoietin (EPO)	18mIU/ml (<25)
24h-urine metanephrines	66 μ g (52-341)
TSH	2.18 μ IU/mL
O ₂ sat	98%

On laparoscopic adrenalectomy, a 2.5 cm adrenal adenoma, Weiss score 0, was removed (Figure 1) without complications and oral hydrocortisone supplementation postoperatively.

One month following excision, her labs were as follows:

hematocrit 42%
hemoglobin 13.6 g/dl
potassium 4.9
testosterone 1.27 nmol/l (0.43-1.24),
EPO 14 IU/l (2.6-34)
DHEAS 7.2 μ g/dl (18.9-205) and undetectable Δ 4-androstenedione.



Figure 1. The adrenal adenoma

Conclusion

- 1) Clinically apparent androgen co-secretion in adrenal Cushing does not necessarily equate to malignancy
- 2) Secondary polycythemia may be a noteworthy multifactorial presentation of a secretory adrenal adenoma independent of smoking status, mediated by direct EPO tumor production and marrow stimulation by cortisol testosterone

References

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