Pheochromocytoma combined with unusual form of Cushing's syndrome and pituitary microadenoma

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Urinary catecholamine levels became normal. Histological examination revealed a typical pheochromocytoma exhibiting a pseudoglandular pattern of growth; the tumor cells were aligned around spaces. Part of the removed tissue displayed a nodule of cortical nodular hyperplasia with both clear and compact cells. ACTH immunohistochemistry revealed no positive cells in the tumor tissue; however, HPLC analysis demonstrated ACTH and various forms of α-melanocyte-stimulating hormone (MSH)-immunoreactive material in the tumor tissue.

Besides pheochromocytomas, extra-adrenal paragangliomas have been associated with Cushing’s syndrome. Immunologic assays of tumor tissues have identified the usual cause as ectopic ACTH production (5, 6). In the present case, the serum ACTH level was normal, suggesting that the pituitary microadenoma might not play a major role in the Cushing’s phenomenon of this patient. Meloni et al. also reported a patient with a benign adrenomedullary tumor associated with bilateral adrenal hyperplasia and Cushing’s syndrome (7).

Our patient, a 44-year-old woman, exhibited moderate obesity, hypertension and psychiatric problems (depression). Her vital signs were: blood pressure (BP) 240/150–160/90 mmHg, pulse rate 100–90/min eurhythmic. Laboratory data showed blood sugar 5.0 mmol/l, and cholesterol 7.1 mmol/l. Serum cortisol levels were normal, but without a diurnal rhythm (02.00 h, 364 nmol/l; 08.00 h, 447 nmol/l). Serum ACTH was in the normal range (13.43 pmol/l). During suppression tests, 2 mg and 8 mg dexamethasone failed to reduce serum cortisol levels. The 24-h urinary epinephrine and norepinephrine levels, as measured by fluorimetry, were 0.106–0.108 and 1.070–0.703 μmol respectively. These elevated values were not suppressed by clonidine. All other hormones measured were in the normal ranges. NMR investigation of the sella turcica revealed a pituitary microadenoma (4 mm in diameter) on the right side of the adenohypophysis.

On abdominal CT, a 2.5 cm mass was seen in the region of the left adrenal. After dibenilyne pretreatment, the left adrenal was removed.

After removal of the left adrenal, the patient displayed clinical and laboratory signs of hypothaladrenism and was treated with glucocorticoid substitutive therapy.

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