Recurrence of adrenal aldosterone-producing adenoma

José María Calvo-Romero, Juan Lucio Ramos-Salado

Summary

Conn’s syndrome (adrenal aldosterone-producing adenoma) and bilateral adrenal hyperplasia are the most common causes of primary aldosteronism. The treatment of choice for patients with aldosterone-producing adenoma is unilateral total adrenalectomy. Recurrence after adequate surgery is exceptional. We present a patient with recurrence of an aldosterone-producing adenoma in the right adrenal gland 9 years after adrenalectomy of an aldosterone-producing adenoma in the same adrenal gland. We conclude that adrenalectomy is not an adequate therapy for patients with adrenal aldosterone-producing adenoma.

Keywords: aldosteronism; adenoma; Conn’s syndrome; adrenalectomy

Hypertension, hypokalaemia, suppressed plasma renin activity and elevated serum aldosterone levels characterise primary aldosteronism. Prevalence estimates for primary aldosteronism vary from 0.05% to 2% of the hypertensive population. Adrenal aldosterone-producing adenoma (APA) and bilateral adrenal hyperplasia (BAH) are the most common causes of primary aldosteronism. The treatment of choice for patients with APA is unilateral total adrenalectomy and for patients with BAH should be pharmacologic. The recurrence of an adrenal APA after adequate surgery is exceptional. We present a patient with recurrence of an adrenal APA after adrenalectomy.

Case report

A 62-year-old woman was admitted to the hospital because of severe hypokalaemia and poorly controlled hypertension. She underwent adrenalectomy for primary aldosteronism due to right APA 9 years ago. The pathological examination revealed a 2-cm diameter adenoma without other abnormalities. After surgery, she had been asymptomatic with normotension and normokalaemia without pharmacologic treatment. In the follow-up, repeated plasma renin activity and serum aldosterone levels were normal. Six months before admission, hypertension resistant to antihypertensive polytherapy, weakness and paraesthesias developed. At entry, her medications comprised fosinopril (20 mg daily), amlodipine (10 mg daily) and hydrochlorothiazide (25 mg daily). There was no family history of hypertension. A physical examination showed only moderate obesity. The blood pressure was 160/110 mmHg. Blood chemistry showed: urea 4.3 mmol/l, creatinine 52 µmol/l, sodium 143 mmol/l, potassium 2.4 mmol/l and chloride 96 mmol/l. Haematology, chest X-ray and electrocardiogram were normal. Antihypertensive therapy was discontinued and potassium chloride supplements were given. On the following days, mild hypokalaemia (3–3.1 mmol/l) persisted, blood pH was 7.50 and HCO₃⁻ 37 mmol/l. The 24-hour urinary potassium was 67 mmol. Free sodium intake, measured supine at 08.00 h before she arose, plasma aldosterone concentration was 3575 pmol/l (normal range 41–412 pmol/l) and plasma renin activity was undetectable. Upright at 08.00 h, plasma aldosterone concentration and plasma renin activity were 2550 pmol/l (137–825 pmol/l) and undetectable, respectively. Plasma aldosterone concentration and plasma renin activity were 2550 pmol/l (137–825 pmol/l) and undetectable, respectively. Plasma aldosterone concentration before and after saline loading was 3230 pmol/l and 3055 pmol/l, respectively. The 24-hour urinary free cortisol was 120 nmol (75–250 nmol). The 08.00 h basal cortisol was 223 nmol/l (140–690 nmol/l). The 08.00 h cortisol after dexamethasone suppression (1 mg at 23.00 h) was 47 nmol/l. The 24-hour urinary free catecholamines, dopamine, epinephrine and norepinephrine were normal. Adrenal computed tomography (CT) showed a 3-cm diameter hypodense nodule without enhancement after intravenous contrast injection in the right adrenal gland and a normal left adrenal gland. Magnetic resonance imaging (MRI) demonstrated the same right adrenal nodule...
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We describe the recurrence of an adrenal APA 9 years after adenomectomy, which is extremely rare12; to our knowledge, only two such cases have previously been reported in the literature.6

The resected adrenal gland sometimes contains macro- or micro-nodular lesions in addition to APA.7 These nodules are multiple and generally bilateral and lead to a more complex differential diagnosis than the presence of a unilateral solitary mass. This bilateral nodularity is not synonymous with hyperplasia, and adrenal venous sampling is required to detect cases of surgically correctable APA.5 The clinical significance of these nodular lesions in patients with APA is not fully clear. They are not thought to contribute to recurrence but to result from intractable hypertension.4 The pathological examination of the adrenal gland in our patient revealed no abnormalities other than the adenoma.

Adrenal scintigraphy with iodo-cholesterol has assisted in distinguishing APA from BAH. Unilateral accumulation of the isotope characterises APA. Adrenal CT is simpler and less expensive than adrenal scintigraphy. In addition, the sensitivity and positive predictive value for APA of scintigraphy and CT are similar, at approximately 70%.14 The sensitivity of scintigraphy is largely dependent on the size of the adenoma, and it is unlikely that diagnostic information will be obtained if findings are normal on the CT scan.7 Adrenal adenomas can be differentiated from metastases by adrenal MRI. A hyperintense rim in T1- and T2-weighted sequences is characteristic for adenomas,8 as in our case. Biochemical tests also aid in differentiating the type of primary aldosteronism. The absence of the normal postural increase in plasma aldosterone concentration supports the diagnosis of APA, with a diagnostic accuracy of 85%.3

The treatment of choice for patients with APA is unilateral total adrenalectomy.1 Some authors debate whether a tumour-bearing adrenal gland should be removed totally or whether adenomectomy is curative. Our patient underwent adenomectomy and recurrence in the same adrenal gland developed 9 years later. We believe that total adrenalectomy should always be performed for APA.

Surgery of APA cures primary aldosteronism, although persistent hypertension, usually mild, occurred in 11–31% of patients.12 Risk factors for persistent hypertension after surgery are age at surgery, poor response to spironolactone treatment, and long duration of hypertension prior to surgery.9 In our case, there was a short history of hypertension and a good response to spironolactone, and hypertension was cured on a 2-year follow-up after second surgery.

Discussion

We describe the recurrence of an adrenal APA 9 years after adenomectomy, which is extremely rare2; to our knowledge, only two such cases have previously been reported in the literature.6

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Figure 2 Adrenal MRI (coronal T2-weighted image): 3-cm diameter isointense nodule without destruction of the gland and with a hyperintense rim (arrow)
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