Case Report

Cushing's syndrome Secondary to Bilateral Functioning Adrenocortical Adenomas

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Case Summary:

A 67 years old lady was admitted with profound weakness and weight loss for 3-4 months. She has background of primary hyperparathyroidism, hypertension and osteoporosis

Examination revealed BP of 175/90 mmHg, slightly plethoric with proximal myopathy and bruising. CTTAP revealed bilateral axillary and para aortic lymphadenopathy and right adrenal adenoma 32x26mm.CT guided biopsy which showed CLL. She was worked up for hypercortisolism.

Table.1 Investigations	Patients values (Normal values)
24 hr urinary free cortisol (1st)	429 (50-300nmol/24hr)
24 hr urinary free cortisol (2nd)	306 (50-300nmol/24hr)
Plasma ACTH (1st)	6 (0.0-40 ng/l)
Plasma ACTH (2nd)	<5 (0.0-40 ng/l)
24 hr urinary Metadrenaline	0.58 (0.0-2.0µmol/24hr)
24 hr urinary Normetadrenaline	0.78 (0.0-4.3µmol/24hr)
Plasma aldosterone concentration	71 (40-310ng/L)
Plasma renin activity	2.8 (3-40mU/L)
Dehyroepiandrosterone sulphate	5.8 (0.26-6.68µmol/L)
Androstenedione	21.2(0.0-3.5nmol/l)

Table:2	Peripheral	Right adrenal vein	Left adrenal vein	A:U Ratio
Baseline Cortisol	475	2559	740	3.45(>2)
Baseline Aldosterone	146	3000	999	
15min Post ACTH				
Cortisol	719	17500	10743	1.62(<2)
Aldosterone	260	14540	10570	
25min Post ACTH				
Cortisol	826	17500	17500	1.0(<2)
Aldosterone	285	13160	14970	

She had an overnight 1mg DST which showed unsuppressed 8am cortisol levels of 156nmol/L. Her 48hr DST results were 793nmol/L to 158nmol/L. She had an MRI adrenals which revealed right adrenal of 3.3cm and left adrenal of 1.4cm (Image 1 and 2) with normal MRI pituitary. DEXA Scan showed osteoporosis. Octreotide scan showed no uptake in both adrenals. Hormonal dynamic studies (AVS) revealed bilateral autonomous secretion of cortisol (Table 2). She underwent laparoscopic bilateral adrenalectomies, commenced replacement therapy with hydrocortisone and fludrocortisone postoperatively. Histopathology showed nodular proliferation of predominantly lipid containing cells suggestive of bilateral adrenal cortical adenomata.

Image 1

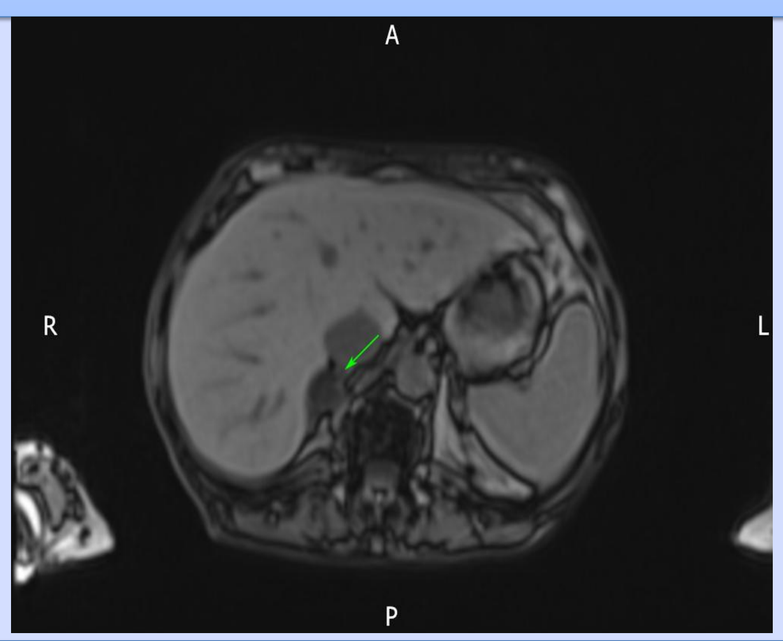
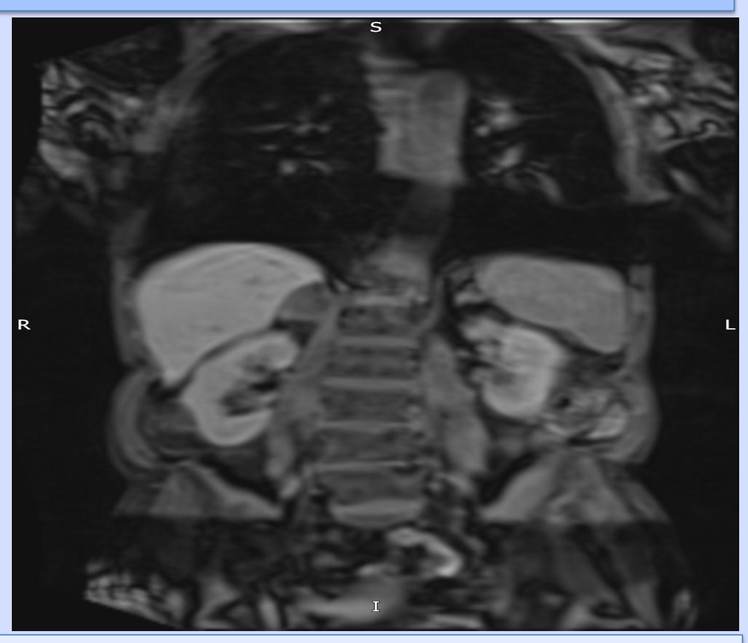


Image 2



Discussion:

Bilateral functioning adrenal adenomas are very rare. Only 25 cases are reported since 1977. Abnormal adrenal expression of receptors for various hormones can lead to ACTH independent bilateral macronodular hyperplasia (AIMAH). These are multiple nodules, not encapsulated with hypertrophic adjacent areas. However cortisol producing adrenal adenomas are usually single, unilateral and encapsulated, usually associated with suppressed ACTH levels leading to atrophy of adjacent non-nodular areas.

References :

1-Iino K, Sasano H, Nagura H, et al. Adrenal adenoma with bilateral adrenocortical nodular change in a patient with Cushing's syndrome. Clin Endocrinol (Oxf) 1997; 47:371.

2-van den Berg G, Frölich M, Veldhuis JD, Roelfsema F. Combined amplification of the pulsatile and basal modes of adrenocorticotropin and cortisol secretion in patients with Cushing's disease: evidence for decreased responsiveness of the adrenal glands. J Clin Endocrinol Metab 1995; 80:3750.