ADRENAL INFARCTION IN ANTIPHOSPHOLIPID SYNDROME DESPITE THERAPEUTIC ANTICOAGULATION

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Introduction:

We report a case of acute adrenal crisis in a patient with the antiphospholipid syndrome in a man who was on a therapeutic dose of warfarin.

Case Description

A 64-year-old man presented with vomiting and abdominal discomfort. Temperature was 37.8°*C*, pulse 85/min and BP 100/63mmHg. On examination, there was generalized hyperpigmentation. He had a history of deep venous thrombosis of the lower limbs on two occasions and was on warfarin. He previously diagnosed primary antiphospholipid syndrome with a strongly positive anticardiolipin IgG antibody. Serum sodium was 127 mmol/l, and serum potassium was 5.7 mmol/l, urea 9.8 mmol/l and Creatinine 103 µmol/l. Haemoglobin was 12.6 g/dl, white cell count of 7.3 x 10^9/l. INR was 3.1 on warfarin, APTT 133 secs and prothrombin 32.7 secs. Primary adrenal insufficiency was suspected. The Tetracosactrin (Synacthen, 250 micrograms) test had a maximum stimulated cortisol of 43 nmol/l (normal > 550 nmol/l) confirming adrenal insufficiency. CT of Abdomen revealed bilateral adrenal enlargement (right adrenal: 3.1 cm X 2.9 cm, left adrenal: 3.0 cm X 2.5 cm). At the time there was diagnostic uncertainty and biopsy considered risky.

Tuberculosis was considered and was treated for a number of months though no bacteriological or other clinical features diagnosed were established. PET scan was negative for evidence of malignancy. Double stranded Anti-Nuclear Antibody was negative. Beta-2 glycoprotein was positive. Adrenal antibodies tested negative. On a repeat CT Abdomen 4 months later, his adrenal glands shrunk to approximately 1.5 cm bilaterally and on review suggested that there was haemorrhagic infarction initially. He made a good recovery on hydrocortisone and fludrocortisone. INR target was increased to 3.5. After 5 year follow up there was no further infarction.

Discussion/Conclusions

This case illustrates that adrenal infarction can occur in antiphospholipid syndrome despite conventional anticoagulation perhaps because of the adrenal vascular has only single venous drainage but multiple arterial arcades making it more susceptible to thrombosis and haemorrhage[1].

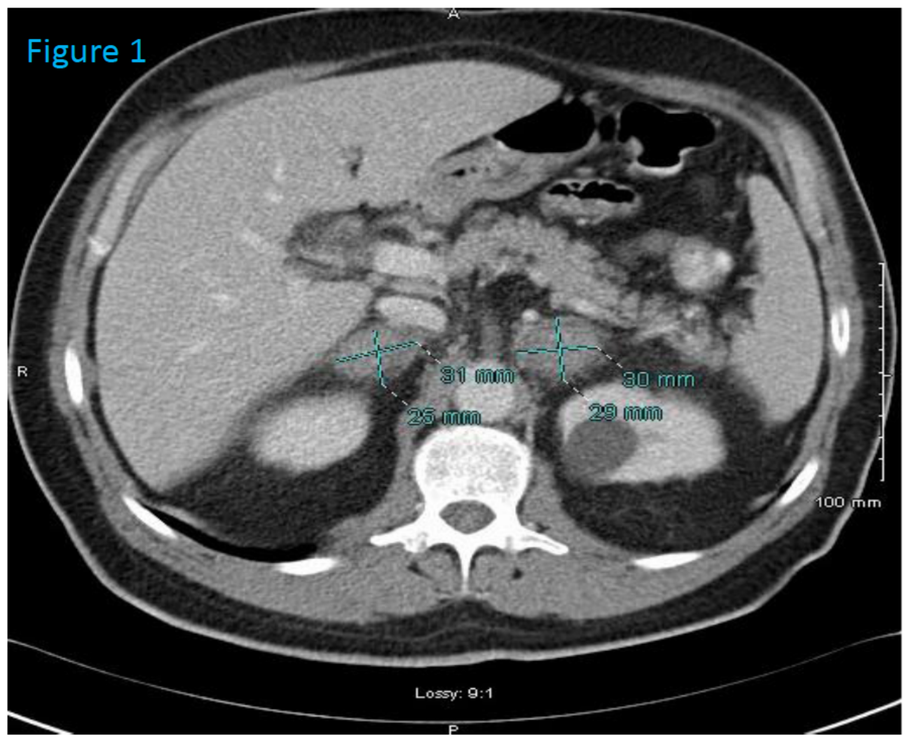

Figure 1:
CT Abdomen at presentation revealed bilateral adrenal enlargement; Right: 3.1 cm X 2.9 cm and left : 3.0 cm X 2.5 cm.

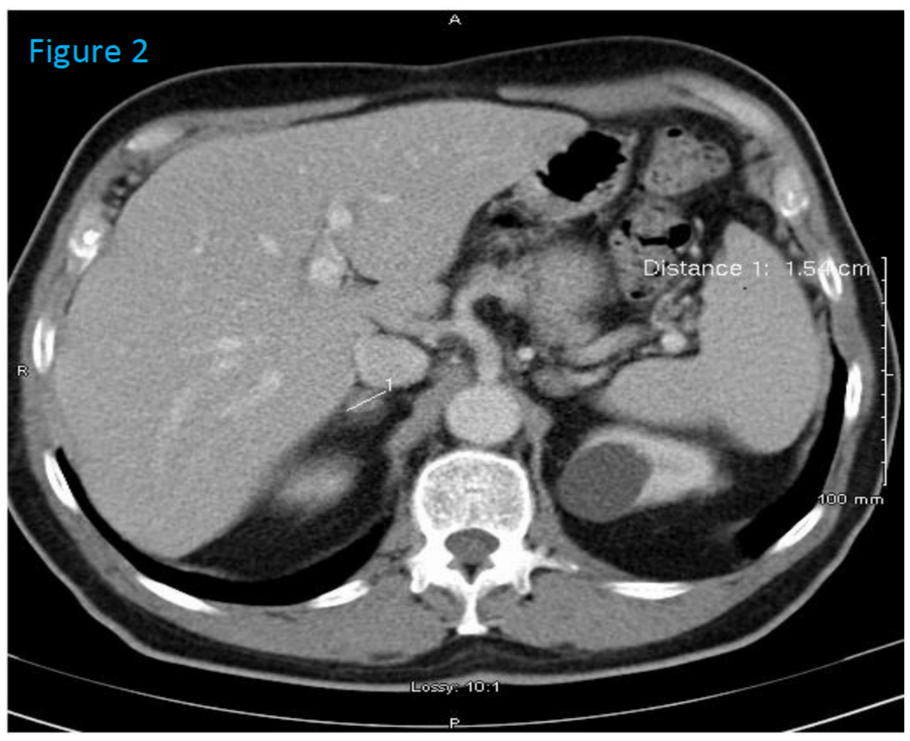

Figure 2:
4 months later, a repeat CT Abdomen demonstrated that the adrenal glands shrunk to approximately 1.5 cm bilaterally.

Reference:
1. Presotto F, Fornasini F, Betterle C, Federspil G, Rossato M. Acute adrenal failure as the heralding symptom of primary antiphospholipid syndrome: Report of a case and review of the literature. Eur J Endocrinol. 2005;153:507–14.