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When a rare syndrome keeps behaving in rarer manners over and over again!

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Pituitary apoplexy arises when haemorrhage and/or infarction occurs within a pituitary tumour. In Malta, the estimated standardised incidence rate (SIR) of apoplexy is 0.15/100,000/yr. ACTH secreting pituitary adenomas have a SIR of 0.17/100,000/year.

Case Report: A 46 year-old gentleman with a history of poorly controlled diabetes mellitus was referred following the diagnosis of a pituitary adenoma. He had presented with a 1.5 year history of left third cranial neuropathy with complete ptosis. Magnetic resonance imaging (MRI) showed a 3.2×1.5 cm lesion extending into the suprasellar cistern, abutting the optic chiasm and extending into the cavernous sinuses, more pronounced on the left side. Biochemical assessment revealed cortisol of 483 nmol/l, prolactin 31 mIU/l, Testosterone 3.5 nmol/l, LH 1.9 U/l, FSH 4.5 U/l, TSH 2.34 micIU/l and T4 12.27 pmol/l. The patient had an inadequately suppressed cortisol level (149 mmol/l) on a 48-hour low dose dexamethasone suppression test and a high ACTH (102 pg/ml) in keeping with ACTH-dependent Cushing's syndrome. The patient was referred for infra-petrosal sinus sampling, which confirmed an ACTH secreting pituitary macroadenoma. The patient could not undergo surgery at the time in view of an infected diabetic foot ulcer. He was initiated on Metyrapone in an effort to control his hypercortisolaemia. The patient presented two months later with severe headache and new onset visual disturbances. He developed a right III and VI cranial nerve (CN) palsies. Ophthalmological assessment revealed a reduction in visual acuity. Areas of hyperintensity in the pituitary adenoma were noted on unenhanced T1 MRI scan and there was lack of enhancement on a contrast scan, in keeping with apoplexy. Urgent debulking was carried out through a trans-sphenoidal approach. Clinical symptoms and visual disturbances showed improvement post-operatively; the right 3rd and 6th CN palsies improved but he had a persistent right temporal visual field defect. Histology confirmed pituitary apoplexy due to tumour infarction of a functional (ACTH secreting) pituitary macroadenoma.

Conclusion: Presentation of Cushing's Disease can be very varied and the work up is extensive and elaborate with a number of different steps. Cushing's syndrome is associated with multiple comorbidities including increased risk of cardiovascular events, neurological consequences osteoporosis and poor quality of life. Hence, it is imperative

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that an early diagnosis is made as early as possible so that the condition is appropriately treated. Although apoplexy is rare, this complication needs to be kept in mind as an inherent risk when managing patients with pituitary adenomas.

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