Unusual presentation of acromegaly and functioning pituitary gonadotrophinoma (FSHoma)

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Background: Gonadotroph adenomas are the most common non-functioning pituitary adenomas in adult. However, functioning gonadotrophinomas are rare. Here, we describe an unusual presentation of concurrent acromegaly and functioning FSHoma (Follicular stimulating hormone-secreting pituitary adenoma).

Clinical presentation:
A 39-year-old man presented with a vague visual disturbance to the optometrist and a bitemporal hemianopia was detected. Further questioning elicited increase in shoe size with no significant history of sweating, headache and change in hand size. Physical examination was notable for features of acromegaly (prominent eyebrows, prognathism, large hands and feet) and bilateral testicular enlargement.

Investigations:
Endocrine profiling confirmed acromegaly on OGTT- oral glucose tolerance test (non-suppressed nadir GH level of 1.2 ug/L with basal GH level of 1.5 ug/L). IGF 1 level was elevated at 64 (9.5-45) nmol/L. There was significantly elevated FSH level of 107.2 (1-10.1) U/L with LH 1.2 (1.5-6.3) U/L and testosterone 9.3 (8-29) nmol/l. The rest of pituitary profiling was normal: Prolactin 370 (45-375) mU/L, TSH 0.65 (0.35 to 5.5) mU/L, FT4 12.3 (10-19.8) pmol/L, normal short synacthen test. US testes confirmed bilateral testicular enlargement: left testis measuring 46cc and right testis measuring 50cc with no neoplastic changes. MRI pituitary demonstrated a large pituitary macroadenoma 3.5x 2.8 cm, with the expansion of pituitary fossa, compressing optic chiasm and extending into left cavernous sinus.

Management:
Subsequently transphenoidal hypophysectomy was performed. Histology confirmed chromophob pituitary adenoma. Immunohistochemistry showed FSH-immunoreactive adenoma with negative GH staining.

Literature analysis revealed very rare synchronous presentation of acromegaly and FSH-secreting pituitary adenoma. Concurrent GH and prolactin secretion is more common.

References: