signs of early recurrence on follow up imaging. **Case:** A 40-year-old man presented with frontal and maxillary sinus pain and nasal stuffiness. Nasal endoscopy revealed a mass in the nasal cavity and sphenoid sinus. Subsequent CT scan of his sinuses and MRI Pituitary reported a 4 x 3.5 x 2.5 cm sellar mass extending through the floor of sella into the sphenoid sinus and nasal cavity. Despite no clinical features of any pituitary hormone excess (including Cushing's) or deficiency, his biochemical parameters were consistent with hypercortisolaemia. Post-low dose dexamethasone (2 mg over 48 hours) suppression cortisol was inadequate: 106 nmol/L (<50nmol/L). His corresponding serum dexamethasone level was 8.6 nmol/L (>3nmol/L), ACTH 46 ng/L (0-46 ng/L) and 24-hour urinary free cortisol levels were 502 and 260 nmol/24 hours (<165 nmol/24 hours)) on 2 separate occasions. The remainder of his pituitary profile was normal. He therefore underwent expanded endonasal endoscopic resection of tumour without significant complication. His post-operative cortisol was <50nmol/L (other pituitary hormone measurements normal) so hydrocortisone replacement therapy was commenced. A 3-month post-operative MRI pituitary scan revealed near total resection of the tumour with peripheral enhancement within the tumour resection site representing post-surgical changes. Histopathology showed sparsely granulated, diffuse cytokeratin immunostaining and immunopositivity for ACTH and T-pit consistent with a corticotroph PitNet of the sparsely granulated variety with low Ki 67 index. His six month post-operative scan showed recurrent sellar mass of 2.2 x 0.9 x 1.4 cm. Again there was an absence of clinical features of Cushing's syndrome. After multidisciplinary discussion clinical, biochemical and radiological surveillance was continued with a view to further transsphenoidal surgery and/or radiotherapy depending on the progress of the tumour.

Conclusion: Our patient solely had biochemical (not clinical) evidence of Cushing's disease despite a large pituitary tumour in keeping with an SCA, which are aggressive, have high recurrence rate and shorter time to recurrence after surgery. Close and frequent post-operative clinical, biochemical and radiological surveillance is critical to detect and manage early recurrence that may require multimodal therapy.

Neuroendocrinology and Pituitary

NEUROENDOCRINOLOGY AND PITUITARY CASE REPORTS

Stuck at the Checkpoint: Pembrolizumab Induced Hypophysitis With 'Normal' Cosyntropin Response

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Introduction: Pembrolizumab is an immune checkpoint inhibitor (ICI) that blocks programmed cell death receptor 1 (PD-1) and is indicated in treatment of malignancies including lung adenocarcinoma and melanoma. Anti-PD-1 therapies are responsible for immune related adverse events including endocrine dysfunction. Here we present a case of Pembrolizumab induced hypophysitis causing central adrenal insufficiency apparently missed by a 250 mcg cosyntropin test.

Clinical Case: A 68 year old female with history of COPD and stage 4 lung adenocarcinoma with bone metastases presented to the ED with hypotension during her 14th cycle of treatment with Pembrolizumab. Other symptoms included fatigue and anorexia for about 4 weeks. She denied vomiting, diarrhea, bleeding episodes or chronic steroid use. On exam, patient was obese and pale appearing, with decreased bilateral breath sounds without edema. Random cortisol on admission at 4:44 pm was 6.9 ug/dl, ACTH - was in process, Na was 135 (137 - 145 mEq/L) and K was 3.7 (3.5 - 5.1 mEq/L). A 250 mcg cosyntropin stimulation test resulted in 30 min cortisol level of 22.5 µg/dL and 60 min cortisol level of 34 µg/dL. Patient was treated with fluids, salt tabs and eventually midodrine, however BP remained borderline low. On day 5, the patient was started on IV methylprednisolone 40 mg Q8 hrs for COPD exacerbation. A few days later pre-steroid ACTH was reported as <5 pg/ml (6-50 pg/ml) which prompted further workup for hypopituitarism. LH and FSH levels were inappropriately low for a postmenopausal female at 0.2 and 2.6 mIU/ml respectively, Free T3 was 2.02 pg/ ml (2.77-5.27 pg/ml), free T4 was 1.07 ng/dl (0.80-2.20 ng/dl), and TSH was 0.67 uIU/ml (0.47-4.70 uIU/ml). MRI brain showed partially empty sella. Patient was diagnosed clinically with Pembrolizumab induced hypophysitis causing central adrenal insufficiency. Eventually, steroids were tapered to prednisone 5 mg daily maintenance dose and patient was discharged with stress dosing instructions.

Conclusion: Diagnosis of adrenal insufficiency is challenging as advanced malignancy and adrenal insufficiency can cause similar symptoms. The finding of an empty sella here is a confounding factor. Checkpoint inhibitors are known to cause hypophysitis, thyroiditis and primary adrenal insufficiency, however incidence is <1%. We report a case of missed adrenal insufficiency by a falsely assuring cosyntropin test. Based on our experience, we conclude that when suspecting a diagnosis of checkpoint inhibitor induced adrenal insufficiency, we should start by checking random cortisol and ACTH value. A standard 250 mcg cosyntropin test should not be used solely to completely rule out this diagnosis.

Neuroendocrinology and Pituitary

NEUROENDOCRINOLOGY AND PITUITARY CASE REPORTS

Successful Multimodal Treatment of a TSH-Secreting Pituitary Adenoma (TSH-oma)

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Background: TSH-omas are rare tumors accounting for 0.5-2% of all pituitary adenomas. Due to their indolent nature, most TSH-omas are diagnosed at the stage of invasive macroadenomas. Over the past several decades, the management of TSH-omas has evolved substantially. While surgery remains first-line therapy, somatostatin analogs have emerged as important therapeutic agents as a result of their effectiveness in normalizing thyroid hormone levels in ~95% of patients with severe hyperthyroidism and reducing TSH-oma size in ~50% of patients.

Clinical Case: A 52-year-old woman with a history of multinodular goiter was incidentally found to have a 2.2 x 1.8 x 2.1 cm pituitary macroadenoma with suprasellar extension, mass effect on the optic chiasm, and left cavernous sinus involvement when she presented with chest pain, palpitations, headache, and left-sided numbness and weakness. Laboratory results showed high FT4/T4/T3 with inappropriately high TSH, elevated α-subunit, and low cortisol with low-normal ACTH highly suggestive of TSH-oma with concurrent secondary adrenal insufficiency. An ophthalmology exam revealed a left superior temporal defect. The patient was treated with atenolol, prednisone, and octreotide two weeks before surgery with symptomatic improvement and near-normalization of FT4. Following an uncomplicated transsphenoidal resection, FT4 normalized within one week. At her one-month follow-up, both TSH and FT4 were normal, and her secondary adrenal insufficiency had resolved. Her visual field defect also recovered.
Laboratory Results: TSH 5.35 (normal range 0.40-4.60 μU/mL), FT4 3.0 (0.8-1.7 ng/dL), T4 18.1 (5.0-12.0 μg/dL), T3 235 (80-200 ng/dL), ACTH 10 (6-50 pg/mL), cortisol 4.5 (5.0-25.0 μg/dL), α-subunit 8.0 (0.1-1.5 ng/mL); after 2 weeks on SQ octreotide 50mg q12h: TSH 1.93 (0.30-4.20 μU/mL), FT4 1.7 (0.6-1.5 ng/dL); 1 month post-op: TSH 1.53 (0.30-4.20 μU/mL), FT4 0.8 (0.6-1.5 ng/dL), ACTH 12 (7.2-63 pg/mL), cortisol 6.9 (4.0-20.0 μg/dL)
Conclusion: Since the first reported case of TSH-oma in 1960, the diagnostic and therapeutic management of these rare pituitary adenomas have evolved due to the emergence of ultrasensitive TSH assays, advanced imaging and surgical techniques, and somatostatin analogs. However, to this day, most TSH-omas are still diagnosed at the stage of invasive macroadenomas, when successful surgical resection becomes more difficult. Hence, up to two-thirds of patients may require adjuvant therapy with medication or radiation. As evidenced in our patient, who achieved a near-euthyroid state within just two weeks of starting low dose octreotide, somatostatin analogs are highly effective in controlling hyperthyroidism and have solidified their place in the therapeutic management of TSH-omas. This case highlights the success of a multimodal approach to the treatment of TSH-omas.

Neuroendocrinology and Pituitary
NEUROENDOCRINOLOGY AND PITUITARY CASE REPORTS

Successful Pregnancies in an Acromegalic Woman After Non-Radical Pituitary Adenomectomy for Somatoprolactinoma

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Background: Pregnancy is unusual in patients with acromegaly due to somatotropinomas or somatoprolactinomas. Fertility is impaired because of hormonal hypersecretion, pituitary damage by tumor compression or both. Managing somatoprolactinomas and fertility issues are often challenging.

Clinical Case: A 20-year woman with primary amenorrhea and headache was diagnosed with hypogonadotrophic hypogonadism secondary to hyperprolactinemia (2500 μg/L, n<23 μg/L). No other abnormalities were found on the pituitary function screening tests. MRI revealed an intra and suprasellar adenoma (2.5x1.8x1.8 cm) with optic chiasm compression. The onset of menses occurred after 11 months under dopaminergic treatment, and tumor size diminished (1.9x1.5x1.5 cm), bringing on optic chiasm decompression. She remained under dopamine agonist treatment for 6 years, when she realized extremities enlargement and height increase by 3 cm. Acromegaly was confirmed by blood levels of IGF-1 (3.37xULN), GH (8 μg/L, n<8 μg/L), and GH nadir (4.3 μg/L, n<1 μg/L) during OGTT. Then, octreotide LAR was added to cabergoline treatment while waiting for elective surgical treatment. She underwent to transsphenoidal endonasal neurosurgical microscopy approach guided by neuronavigation, with the removal of a large portion of tumor. However, it was not possible to extract the part of invasive adenoma close to right carotid artery due to the risk of vascular and intracavernous cranial nerves injury. Immunohistochemistry analysis of the adenoma was positive only for GH cells with low Ki67 index (<1%). Due to the poor biochemical control (unsuppressed post-OGTT GH, IGF-1 1.66xULN and PRL 301 μg/L) and the presence of a small stable tumor residue, treatment with cabergoline and somatostatin analogues was maintained (3-year octreotide LAR, transitioned to lanreotide in an attempt to achieve a better biochemical response). After 14 years of the initial diagnosis and 5 years post-surgery, the patient expressed the desire to get pregnant and all medications in use were suspended. In the following 3 years, she had two uneventful gestation without complications or worsen of acromegaly; she only breastfed for few months after her first pregnancy. The second one was a twin pregnancy. After one year, the MRI revealed no increase of tumor mass (1.0x0.3x1.0 cm), and PRL levels withing normal range, IGF-1 slightly elevated, but GH not suppressed by OGTT. Cabergoline was reintroduced and the biochemical control of acromegaly was achieved.
Conclusion: We reported the very unusual spontaneous conception and normal course of pregnancies in a woman with acromegaly, who was submitted to successful transsphenoidal neurosurgical microscopy approach in which large part of the tumor was removed and the normal pituitary tissue was preserved, allowing fertility restoration.

Neuroendocrinology and Pituitary
NEUROENDOCRINOLOGY AND PITUITARY CASE REPORTS

Successful Treatment of Myxedema Coma Using Liothyronine in the Setting of Adrenal Crisis

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Background: Myxedema coma (MC) represents severe decompensated hypothyroidism and is associated with