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 transphenoidal 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 μg/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-old woman with primary amenorrhea and headache was diagnosed with hypopituitary hy- pogonadism 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 transphenoidal 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 re- sponse). 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 transphenoidal 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 Lithrothine in the Setting of Adrenal Crisis

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Background: Myxedema coma (MC) represents severe decompensated hypothyroidism and is associated with
A 58-year old man presented for evaluation of acromegaly. He described symptoms of increased sweating, peripheral glucose use; however, hypoglycemia related to acromegaly has not been reported.

Clinical Case: A 58-year old man presented for evaluation of several elevated serum IGF1 levels. The patient had reported years of increased body heat but no changes in his hands or feet and no voice deepening. He recently needed 15 dental crowns due to gaps in his teeth. He also had difficult to manage OSA and weight gain. The patient reported neuroglycopenia after a high glycemic meal or drink, although he was never able to objectively measure any low blood sugars when they occurred; these symptoms improved but did not resolve despite adhering to a low carbohydrate diet. He also had decreased libido and erectile dysfunction. Exam was significant for coarse facial features. Prior testing revealed several elevated IGF1 serum levels, the last one being 227 ng/mL (54-194). One year prior, OGTT resulted in an initial GH level of 0.1 ng/mL with a decrease to <0.1 ng/mL after two hours. Repeat OGTT had an initial GH of 2.98 ng/mL which paradoxically rose to 12 ng/mL. Fasting BG was 90 mg/dL and peaked at 171 mg/dL. Pituitary MRI showed a 5 mm microadenoma, consistent with acromegaly from a GH secreting adenoma. He underwent a TSSC, and his heat intolerance, low libido, and symptom of hypoglycemia resolved completely. Subsequent IGF1 levels and MRI imaging normalized. Postoperatively OGTT showed a peak GH of 0.23 ng/mL with a peak glucose of 134 mg/dL. There was no paradoxical rise in GH.

Discussion: Acromegaly is commonly associated with insulin resistance in ~30% of cases; however, there are no reports of associated neuroglycopenia after a carbohydrate-rich meal or OGTT, which in our patient resolved after successful removal of the pituitary microadenoma. His low glucose symptoms could have been a result of reactive hypoglycemia, which is often seen in patients with diabetes or even prediabetes. However this patient had no history of either. He did not have evidence of any tumors causing hypoglycemia and no gastric surgery to suggest a related etiology (e.g. dumping syndrome or nesidioblastosis). Conversely since GH is normally anabolic and stimulates insulin release, the patient’s elevated GH may have caused an abnormal increase in insulin, leading to his hypoglycemia symptoms. Indeed GIP, which stimulates insulin, is thought to be the cause of the paradoxical rise in GH seen in 30% of acromegaly cases. Remarkably, the patient’s hypoglycemia symptoms disappeared after treatment of the acromegaly, which leads us to consider that excess GH was the culprit.

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NEUROENDOCRINOLOGY AND PITUITARY CASE REPORTS

Surgical Remission of Acromegaly Resolves Neuroglycopenia and Paradoxical Rise in GH after OGTT

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NEUROENDOCRINOLOGY AND PITUITARY CASE REPORTS

The Biggest Man in the Room
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A 42 year old gentleman who had been healthy all his life, began to develop new clinical symptoms including acid reflux. He was tested for H. Pylori by his PCP, and...