Crooke Cell Adenoma as a Rare Cause of Recurrent Cushing Disease: A Challenge in Treatment and Follow-up

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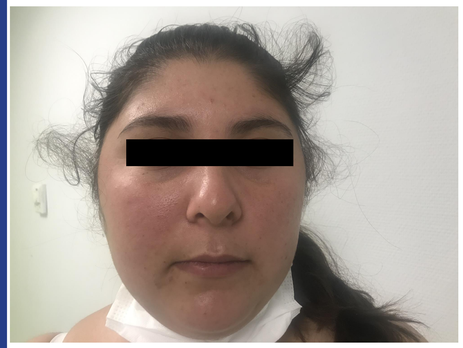
INTRODUCTION

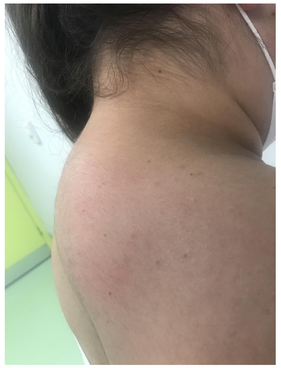
Crooke's cell adenoma (CCA) is an aggressive subtype of corticotroph adenoma, it is usually large,frequently invades surrounding tissues, and is resistant to both surgery and radiation therapy.

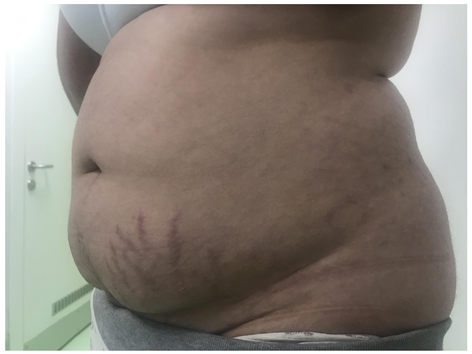
CASE

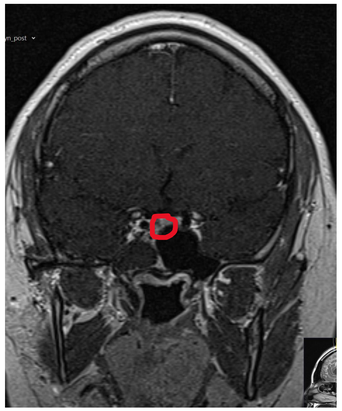
We herein report a case that needed reoperation due to Crooke cell Cushing's disease.

Case Report: A 13-year-old girl, admitted with weight gain for more than 3 months. Her height was 147.5 cm (-1.5 SDS), weight 58.5 kg (1.1 SDS), BMI 26.89 kg/m^2 (2 SDS) and blood pressure was 100/78mmHg. Her physical examination revealed truncal obesity, buffalo hump, moon-shaped face, acanthosis nigricans, abdominal striae and Tanner stage IV puberty. Early morning fasting blood glucose, lipids and thyroid function tests were normal, but serum cortisol was elevated and had lost its diurnal rhythmicity. 24-hour free urine cortisol level was high: 193 ug / L (4-56). The serum cortisol level was not suppressible (20.1 µg/dL) by low-dose dexamethasone, but was suppressed to 1.09 µg/dL by high-dose dexamethasone test. A pituitary magnetic resonans imaging (MRI) did not reveal a discrete adenoma and bilateral inferior petrosal sinus sampling showed that the pituitary was the source of Cushing syndrome. The inter petrosal sinus ACTH gradient indicated ACTH to the right side. Transsphenoidal right-sided pituitary adenomectomy was performed, and pathology result revealed Crooke cell adenoma. Postoperatively she had a normal pituitary reserve, with the exception of hypothyroidism and was commenced on thyroxine therapy. Three years later, she re-presented with rapid weight gain, menstrual cycle irregularity. In this time,her weight was 82.8 kg (3.0 SDS), height 156.6 cm (-1.09 SDS) and BMI was 33.7 kg/m^2 (3.2 SDS). Early morning ACTH and cortisol levels were 51.8 pg / m L and 23.9 µg/dL respectively. Her 24-hour urinary free cortisol level was 46.26 ug / L (3-43). MRI examination revealed postoperative changes and 5x5 mm adenoma on the right hemisphere of the hypophysis. Cabergoline treatment was initiated for the patient. However, there was no response to the treatment within 6 months and the second transsphenoidal resection was performed.









CONCLUSIONS

These tumors are usually invasive, may exhibit aggressive clinical behavior, and often recur with a low success of cure after reoperation and/or radiotherapy. These tumors may betreated by several treatment modalities and recent work has elucidated new targets for more effective elimination andremission.

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