Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Endocrinology

A Rare Case of Plurihormonal Pituitary Adenoma Associating Cushing's Disease and Silent Prolactinoma

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DOI: 10.36347/sjmcr.2022.v10i05.001 | **Received:** 26.03.2022 | **Accepted:** 02.05.2022 | **Published:** 07.05.2022

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Abstract | **Case Report**

Plurihormonal pituitary adenomas are rare conditions that secrete two or more pituitary hormones. The association of prolactin with other hormones is described but associated with ACTH is uncommon. We report a rare case of mixed adenoma associating Cushing's disease and silent prolactin adenoma in 21 years old male patient. This case highlights an unusual plurihormonal pituitary adenoma case with a rare association between ACTH and prolactin. It pathogeny is less clearly define.

Keywords: Plurihormonal pituitary adenoma, Cushing's disease, prolactin.

INTRODUCTION

Plurihormonal pituitary adenomas are rare condition that secretes two or more pituitary hormones. The association of prolactin with other hormones is described but associated with ACTH is uncommon. We report a rare case of mixed adenoma associating Cushing's disease and silent prolactin adenoma.

CASE

It's a 21 year-old male patient with prior history of hypertension controlled by therapy of 3 pills per day and a pre-diabetes under hygiene and dietary measures. He came complaining of obesity with body changes and bone pain since few years. He reports a decrease in morning erections without an intracranial hypertension syndrome or galactorrhea.

Clinical examination showed Cushing features with moon face, facio-truncular distribution of fat and slender limbs, purple stretch marks on the abdomen, internal surfaces of the legs and armpits and with several statutory delay and no gynecomastia or galactorrhea (figure1). He has adult-looking external genitalia with micropenis and 10-12 mL orchidometer.

The positive diagnostic of Cushing disease was deal by urinary free cortisol (UFC) test 5 times the upper limit and a negative overnight 1mg dexamethasone suppression test, a high ACTH level at 211,50pg /ml and confirmed by hypothalamic-pituitary MRI with microadenoma measuring approximately 3.3*4.6 mm lateralized to the right with median pituitary stalk.(figure 2) The hormonal tests showed normal prolactina at 13,74ng/ml, gonadotropic deficiency, normal thyrotropic axis and IGF-1 wasn't asked. A left ventricular hypertrophy is noted on the EKG and osteoporosis is found on the bone densitometry.

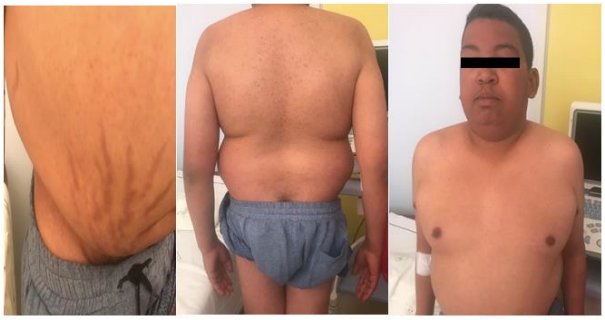

Fig-1: Cushing's fealures on examination

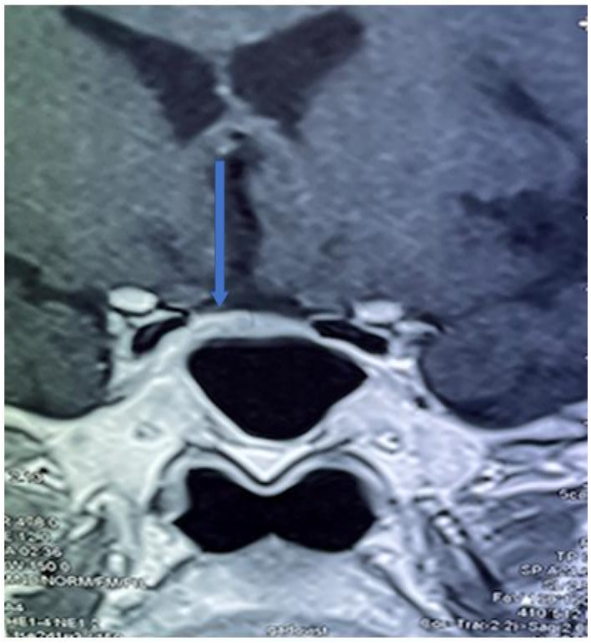

Fig-2: Preoperative coronal hypothalamic-pituitary MRI showing a microadenoma measuring approximately 3.3*4.6 mm lateralized to the right

The transsphenoidal pituitary surgery was performing with simple surgical follow-up without signs of adrenal insufficiency with cortisol at 34,6µg/dl. The absence of remission was confirmed with upper UFC done a post after the surgery at 596µg/24h (4N) and post-operative prolactinoma still in normal value.

The anatomopatho-immunohistochimical study concludes to a morphological aspect of pituitary adenoma with diffuse granular cytoplasmic expression of 60% of anti-prolactin antibody tumor cells and low to moderate cytoplasmic expression of anti-ACTH antibody tumor cells with Ki67 at 3%.

The management of our patient will depend of clinical evolution of the disease and post-operative MRI. At this time clinically he losed weight, then the hypertension treatment was reduce to one pill per day. We started to treat the gonadotropic deficiency with injectable testosterona.

DISCUSSION

Plurihormonal pituitary adenomas are a unique type of pituitary adenomas that secrete two or more pituitary hormones normally associated with separate cell types that have different immunocytochemical and ultrastructural features. The most common hormone combinations secreted by plurihormonal pituitary adenomas are growth hormone, prolactin, and one or more glycoprotein hormones [1]. In our case it's about a rare association between Cushing's disease and silent lactotrophic pituitary adenoma.

Mixed ACTH and PRL adenomas are often manifested by the coexistence of the Cushing's disease and prolactinoma [2]. The clinical presentation of our patient was the same of typically Cushing's disease by microadenoma without any signs of lactotrophic's expression in male patient as gynecomastia.

The pathogenesis of plurimorphous plurihormonal tumors is less clearly defined, but it has been suggested that it may result from neoplastic transformation of two separate cell lines or from the transdifferentiation of one single tumor cell line into a different hormone- producing cell line [3]. In fact, in humans the genes for PRL and ACTH production are too different to be simultaneously expressed in the same cell suggesting that these adenomas can only contain distinct tumor cell lineages, in the sense that two distinct tumors growing in a small space could join together in a single mass [4].

The statural delay reflects the impact of Cushing's disease in bone maturation, thus generating a short final height. Glucocorticoid suppressed GH-induced secretion and action of IGF-1 in rat chondrocytes [5]. High cortisol levels may also reduce the number and function of mature osteoblast cells by interfering with the differentiation and replication. It also induces apoptosis of osteoblasts and osteocytes [6].

All criterias of Cushing's disease was founded in our patient and we performed a goal treatment. The initial treatment of choice is pituitary surgery, usually consisting of selective removal of the pituitary tumor (adenomectomy) [7]. There wasn't a surgical remission in our case.

According of the immunochemical expression prolactin antibodies at 60% an adjuvant therapy with dopamine agonist after clinical evaluation and the result of post-operative MRI could be done. Leah T. Braun et al. reported a follow-up study of 20 patients operated on for Cushing's disease, some of whom co-expressed ACTH and PRL on immunohistochemistry, and showed a possible therapeutic benefit with dopamine agonists [8].

CONCLUSION

This case highlights an unusual plurihormonal pituitary adenoma case with a rare association between ACTH and prolactin. It pathogeny is less clearly define.

REFERENCES

1. Allehaibi, E., AlMalki, M. H., & Brema, I. (2021). Plurihormonal pituitary macroadenoma: a case report. *Journal of medical case reports*, *15*(1), 1-6.
2. Dai, C., Kang, J., Liu, X., Yao, Y., Wang, H., & Wang, R. (2021). How to Classify and Define Pituitary Tumors: Recent Advances and Current Controversies. *Frontiers in endocrinology*, *12*, 229.
3. Kannan, S., Staugaitis, S. M., Weil, R. J., & Hatipoglu, B. (2012). A rare corticotroph-secreting

tumor with coexisting prolactin and growth hormone staining cells. *Case reports in endocrinology*, *2012*.

4. Huan, C., Lu, C., Xu, G. M., Qu, X., & Qu, Y. M. (2014). Retrospective analysis of Cushing's disease with or without hyperprolactinemia. *International Journal of Endocrinology*, *2014*.
5. Jux, C., Leiber, K., Hügel, U., Blum, W., Ohlsson, C., Klaus, G., & Mehls, O. (1998). Dexamethasone impairs growth hormone (GH)-stimulated growth by suppression of local insulin-like growth factor (IGF)-I production and expression of GH-and IGF-I-receptor in cultured rat chondrocytes. *Endocrinology*, *139*(7), 3296-3305.
6. Baron, J. E. F. F. R. E. Y., Huang, Z., Oerter, K. E., Bacher, J. D., & Cutler Jr, G. B. (1992). Dexamethasone acts locally to inhibit longitudinal bone growth in rabbits. *American Journal of Physiology-Endocrinology and Metabolism*, *263*(3), E489-E492.
7. Pivonello, R., De Leo, M., Cozzolino, A., & Colao, A. (2015). The treatment of Cushing's disease. *Endocrine reviews*, *36*(4), 385-486.
8. Braun, L. T., Rubinstein, G., Zopp, S., Vogel, F., Schmid-Tannwald, C., Escudero, M. P., ... & Reincke, M. (2020). Recurrence after pituitary surgery in adult Cushing's disease: a systematic review on diagnosis and treatment. *Endocrine*, *70*(2), 218-231.