CYCLIC CUSHING'S SYNDROME – A DIAGNOSTIC CHALLENGE

Daniela Magalhães[1,2,3], Rita Bettencourt-Silva[1,2,3], Joana Queirós[1,3], Paula Freitas[1,2,3], Eduardo Vinha[1,3], Duarte Pignatelli[1,3], Josué Pereira[4], Davide Carvalho[1,2,3]

[1]Endocrinology, Diabetes and Metabolism Department of Centro Hospitalar de São João, Porto, Portugal [2]Faculty of Medicine, University of Porto, Portugal [3]Instituto de Investigação e Inovação em Saúde, University of Porto, Portugal [4]Neurosurgery Department of Centro Hospitalar de São João, Porto, Portugal

SÃO JOÃO HOSPITAL

INTRODUCTION

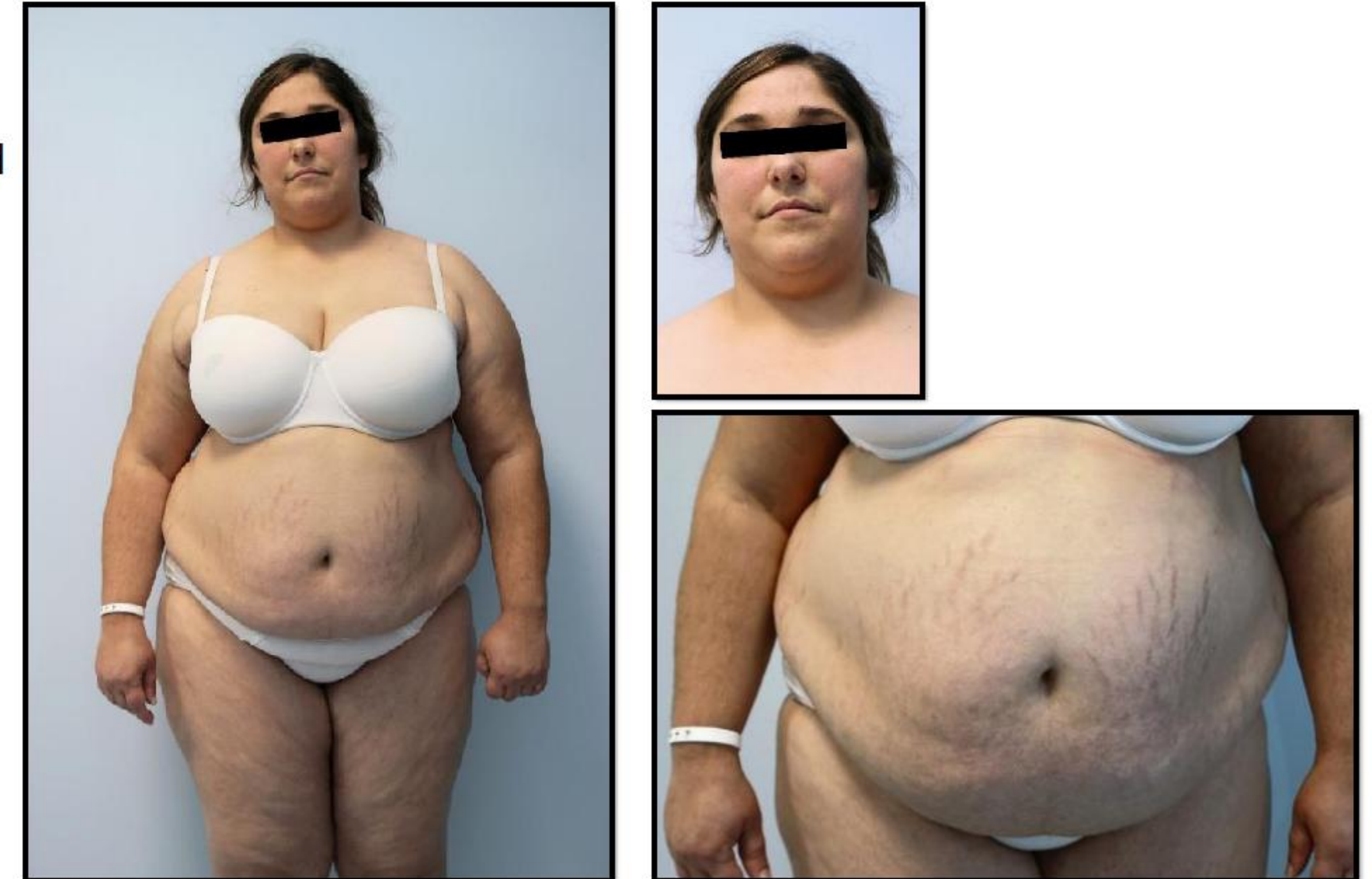
- The diagnosis of Cushing's syndrome (CS) is often challenging considering that none of the used laboratory tests has an ideal diagnostic accuracy.
- The complexity of the diagnosis increases in cyclic disorder, which is characterized by repeated episodes of cortisol excess followed by periods of normal cortisol secretion.

CASE REPORT

32-year-old woman

History of arterial hypertension

Medication: olmesartan and hydrochlorothiazide 20/25mg qd, bisoprolol 10mg qd, amlodipine 10mg qd

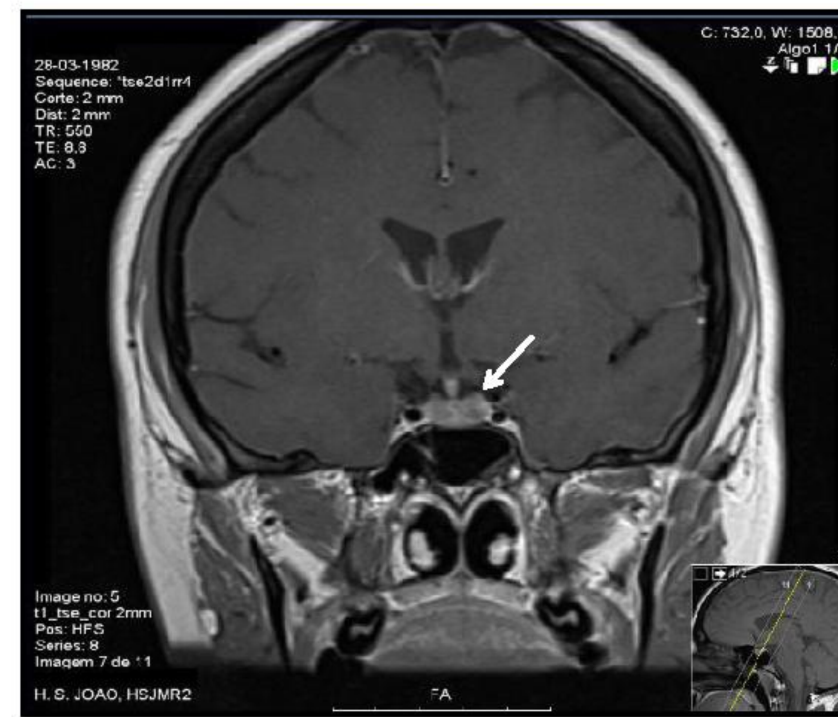


2014

- Weight gain of ≈20kg in 3-4 months with central predominance, dorsocervical fat pad and supraclavicular fullness;
- Facial plethora, facial fullness and acne;
- Abdominal and axilar reddish purple striae;
- Easy bruising;
- Proximal muscle weakness;
- Hirsutism;
- Peripheral edema.

Jun/2014

	Result	Normal Range
Cortisol (8h)	**33.8** µg/dL	6.2-19.4
Cortisol (16h)	**31.7** µg/dL	2.3-11.9
ACTH (8h)	**52.5** ng/L	<63.3
ACTH (16h)	**29.6** ng/L	
Overnight 1-mg DST	**24.3** µg/dL	<1.8
24-h UFC	**930.0** µg/dia	36-137



Pituitary MRI: In the lateral aspect of the left lobe of adenohypophysis is identified a lesion with ≈ 4 mm. There's no involvement of the stalk or the ipsilateral cavernous sinus. It most likely corresponds to a pituitary microadenoma.

Aug/2014

Overall improvement of symptoms

48-hours 2mg/day low-dose dexametasone test showed a final cortisol level of 1.3 µg/dL (<1.8).

Feb/2015

Relapsed symptoms.

Several samples of late-night salivary cortisol revealed mixed results: **0.680** / 0.223 / **0.395** / **1.680** µg/dL (<0.32).

A new pituitary MRI showed a slight increase in the aforementioned area, now measuring ≈ 5.3 mm.

Jul/2015

	Cortisol 8h (µg/dL)	Cortisol 16h (µg/mL)	ACTH 8h (pg/dL)	ACTH 16h (pg/mL)	24-h UFC (µg/day)
Basal	20.0	33.7	7.5	90.7	880.6
Low-dose	**29.5**		74.7		166.0
High-dose	**16.3**		**68.5**		

48-hours low-dose dexametasone suppression test showed a final plasma cortisol level of 29.5 µg/dL, and then a high-dose dexametasone supression test showed a final cortisol level of 16.3 µg/dL, confirming ACTH-dependency (68.5 pg/ml).

Nov/2015

The patient underwent transsphenoidal surgery to remove the pituitary lesion.

Anatomopathological analysis revealed pituitary tissue with no significant changes.

CONCLUSION

This case report documents a cyclic Cushing's disease. The duration of the periods of normal and abnormal cortisol secretion can vary significantly, so the correct diagnosis can be a challenge in clinical practice. Given the possible variations in steroidogenesis, the clinical and biochemical response to medical treatment and the results of drug studies and surgery in cyclic CS must be interpreted with caution. Management of these patients is complicated by high recurrence rates and low cure rates, which necessitate the need for continued closer monitoring for longer durations of time than for patients with noncyclic CS.

BIBLIOGRAPHY: Bonert V, Bose N, Carmichael JD. Cyclic Cushing's disease with misleading inferior petrosal sinus sampling results during a trough phase. Neurosurg Focus. 2015 Feb;38(2):E7. Albiger NM, Scaroni CM, Mantero F. Cyclic Cushing's syndrome: an overview. Arq Bras Endocrinol Metabol. 2007 Nov;51(8):1253-60. Review. Farage M, Costa MA, Godoy-Matos AF. A rare case of Cushing syndrome by cyclic ectopic-ACTH. Arq Bras Endocrinol Metabol. 2012 Jul;56(5):324-30. .

PosterSessionOnline.com