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ADRENAL EMBOLIZATION IN SEVERE ECTOPIC CUSHING: UNUSUAL CASE, EXTRAORDINARY MEASURES

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Introduction

Pancreatic ACTHomas have a poor prognosis with severe and rapidly progressive clinical courses, influenced by hypercortisolemia thus whenever possible, control of the cortisol levels should be obtained to reduce complications. We report the use of bilateral adrenal embolization in a case of a life threatening paraneoplastic Cushing.

Case Report

A 51 year old woman with a negative past medical history complained of general weakness.

The diagnosis of Cushing Syndrome was suspected on the basis of hypokaliemia, new onset hyperglucemia and metabolic alkalosis found in the laboratory tests run on admission. She had no Cushingoid features except mild plethora. The suspicion was confirmed by high levels of urinary free cortisol, serum cortisol and ACTH, a lack of circadian rhythm of serum cortisol, and a low(1mg) and high dose(8mg) overnight of dexamethasone tests that failed to suppress. CT and whole body PET scan showed 2x3 cm mass in the pancreas tail. She underwent the excision of the pancreatic mass and pathology confirmed the neuroendocrine nature of the tumor.

	Plasma cortisol(μg/dl) (4,2-22)	Plasma ACTH(pg/ml) (10-52)	U- cortisol μg/24h
Basal	102,6	530	9800
OVERNIGHT 1mg Dexamethasone suppression test	129,8		
Midnight cortisol	132,5	427,3	
Low dose Dexamethasone suppresion test	146	586,5	
High dose dexamethasone suppression test	141,2	577,8	23247

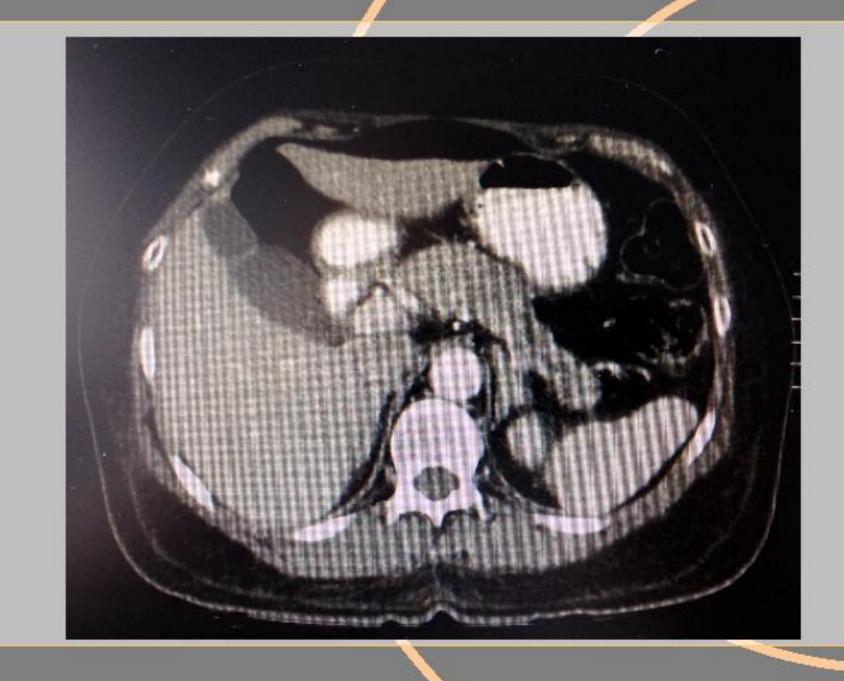
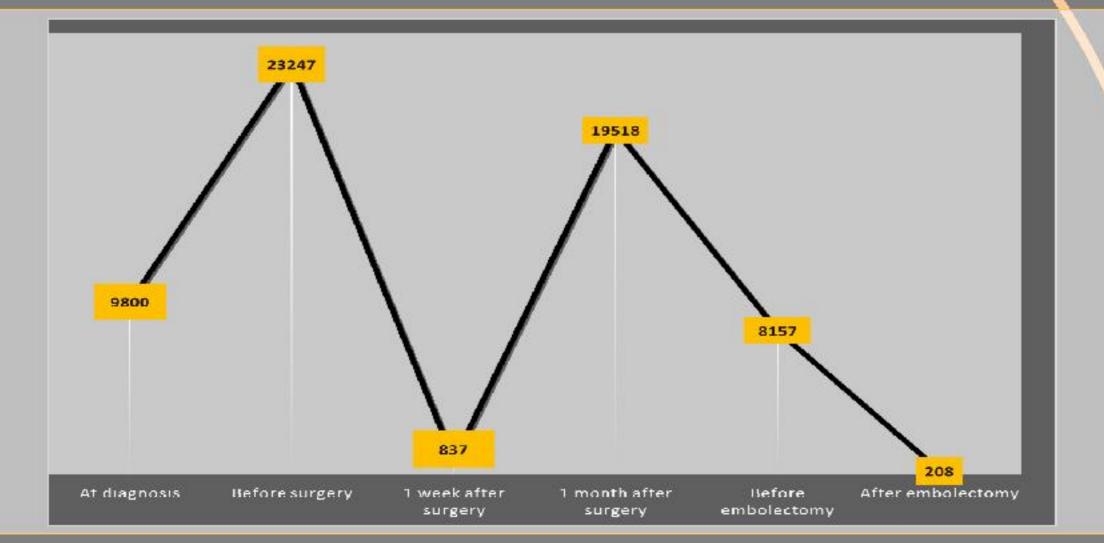


FIG 1: The CT scan shows a 2x3cm mass in the tail of the pancreas, and an important bilateral adrenal enlargement

Few days after the surgery, Cushingoid features were more obvious and liver metastases were observed so treatment with ketokonazol was started, initially decreasing cortisol levels, but refractory hypercortisolemia recurred although dose was increased and metopirone added.

The patient became psychotic so mifepristone was commenced, but she continued to deteriorate. It was therefore decided that an attempt should be made for bilateral adrenalectomy, but the patient was unfit for surgery so a bilateral adrenal embolectomy was performed, in an attempt to lower the cortisol levels and gain some time for the medical treatment to be more effective and to be able to perform adrenalectomy once the patient's condition improved.

Evolution of the urinary cortisol levels



A week after the procedure her condition improved and the urinary cortisol levels lowered. Unfortunately the patient died soon after due to a multiorganic failure.

Conclusions

Aggressive attitudes to control cortisol levels are necessary to reduce comorbilities in Ectopic Cushing. Treatments include excision of the primary tumor, bilateral adrenalectomy and medical control of cortisol levels. This case shows that adrenal embolectomy might be of some use when the patient has become unfit for surgery and in whom medical therapy has failed.

References

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