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**Objective:** Cabergolin is a long acting dopamine receptor agonist used for the treatment of patients with recurrent Cushing's disease.

**Case:** 16 year-old female patient admitted to the hospital because of short stature, amenorrhea, facial and body hair growth, rapid weight gain, hair loss and excessive acne. Her birth weight was 3650 gr, neuromotor development was normal.

#### The first reference(15 years):

- Short stature, amenorrhea, facial and body hair growth

#### At the time of diagnosis (16 years)

- Weight 50 kg (- 0.7 SDS)
- Height 138 cm (-4.0 SDS)
- BMI 26.2 kg/m<sup>2</sup> (+1.7 SDS)
- **Blood Pressure: 145/95 mmHg**
- (Hypertensive)
- A3 P5 M5 (**primer amenorrhea**)
- **Akantosis nigrikans**
- **Buffalo hump**
- **Hirsutismus**
- **Purple striae**
- **Cushingoid appearance**
- **Pletore**
- **Central obesity**

#### Laboratory findings:

- CBC, lipids
- LH, FSH, E2 17-OH-P
- Pelvic USG, BMD
- **Free testosterone: 3.7 pg/ml**
- **ABPM- non-dipper HT**  
(ACE inhibitor treatment )

NORMAL

Table1. ACTH-Cortisol Diurnal rhythm

	Morning	Midnight
Cortisol (μg/dl)	20	17
ACTH	60	52

Table 2. Dexamethasone suppression test

	Suppression test	Low-dose suppression.	High-dose suppression
Cortisol (μg/dl)		27	0.64
ACTH		68	19

#### Pituitary MRI: no pituitary adenoma

Table 3. Petrosal Sinus Sampling

Petrosal Sinus Sampling	Petrosal sinus - left	Petrosal sinus right	Periphery blood
ACTH pg/ml	215	115	29.4
ACTH 5 min	>1250	725	68.1
ACTH 10 min	>1250	>1250	333

\*\* ACTH hypersecretion and lateralization to the left was found by petrosal sinus sampling.

## Cushing's Disease

#### First operation (16,5 years)

Endoscopic transnasal hypophysectomy  
(not successful)

Morning ACTH : 60,4 pg/ml Cortisol : 20 μg/dl  
Midnight ACTH: 52,5 pg/ml Cortisol : 17,14 μg/dl

#### Second operation (17 years)

Endoscopic transnasal left hemi-hypophysectomy  
Clinical findings not progress and the patient lost weight,  
markers of insulin resistance regressed

Morning ACTH: 38,5 pg/ml Cortisol : 10,18 μg/dl  
Midnight ACTH: 37,9 pg/ml Cortisol : 9,41 μg/dl

\*\* Urinary free cortisol: 70 μg/day (Normal)

PATOLOGY : no findings belonging to the adenoma

15 months after second operation → Cushing's disease relapsed clinically and biochemically

Morning ACTH: 69,9 pg/ml Cortisol : 17,7 μg/dl  
Midnight ACTH: 76,6 pg/ml Cortisol : 10,65 μg/dl

\*\* Urinary free cortisol: 197,25 μg/day (N: 36-137 μg/day )

**Conclusion:** Cabergoline is effective in the control of cortisol secretion in the treatment of recurrent Cushing's disease. Diagnosis and treatment of Cushing's disease in children are challenging.

## Recurrent Cushing's Disease after pituitary surgery

- Mortality and morbidity risk of the third operation.
- Radiation therapy – risk of develop panhypopituitarism
- Bilateral adrenalectomy - Nelson syndrom and the risk of severe complications
- Medical treatment ?

**Cabergolin tb**  
**1 mg / week**  
(18,5 years)

After 9 months → Cushing's disease regressed clinically and biochemically  
**Cabergolin treatment**

- Weight 44 kg (- 1,8 SDS)
- Height 143 cm (-2,73 SDS)
- BMI 21.2 kg/m<sup>2</sup> (-0,07 SDS)
- ACTH: 33 pg/ml (N: 10-46 pg/ml)
- Cortisol: 7,32 μg/dl (N: 6,7-22,6 μg/dl)
- Urinary free cortisol: 8,82 μg/day (N: 36-137 μg/day)

Figure 1a.



Figure 1b.



Figure 1 a and b. Before Cabergolin treatment

Figure 2. After Cabergolin treatment

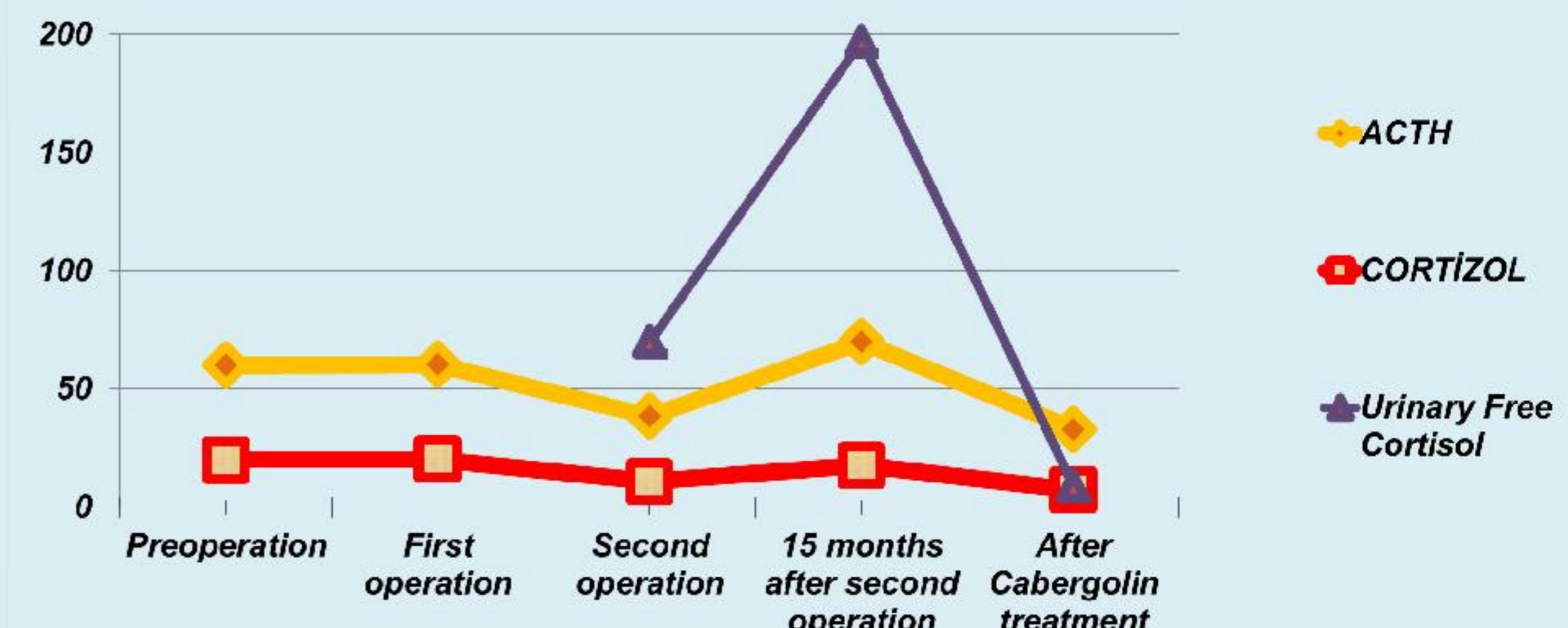


Figure 3: Cases of level ACTH, cortisol and urinary free cortisol in preoperation, after first operation, after second operation, 15 months after second operation and 9 months after cabergolin treatment

