

## Long term follow-up of a child treated with CyberKnife radiosurgery for ACTH-secreting pituitary adenoma after bilateral adrenalectomy



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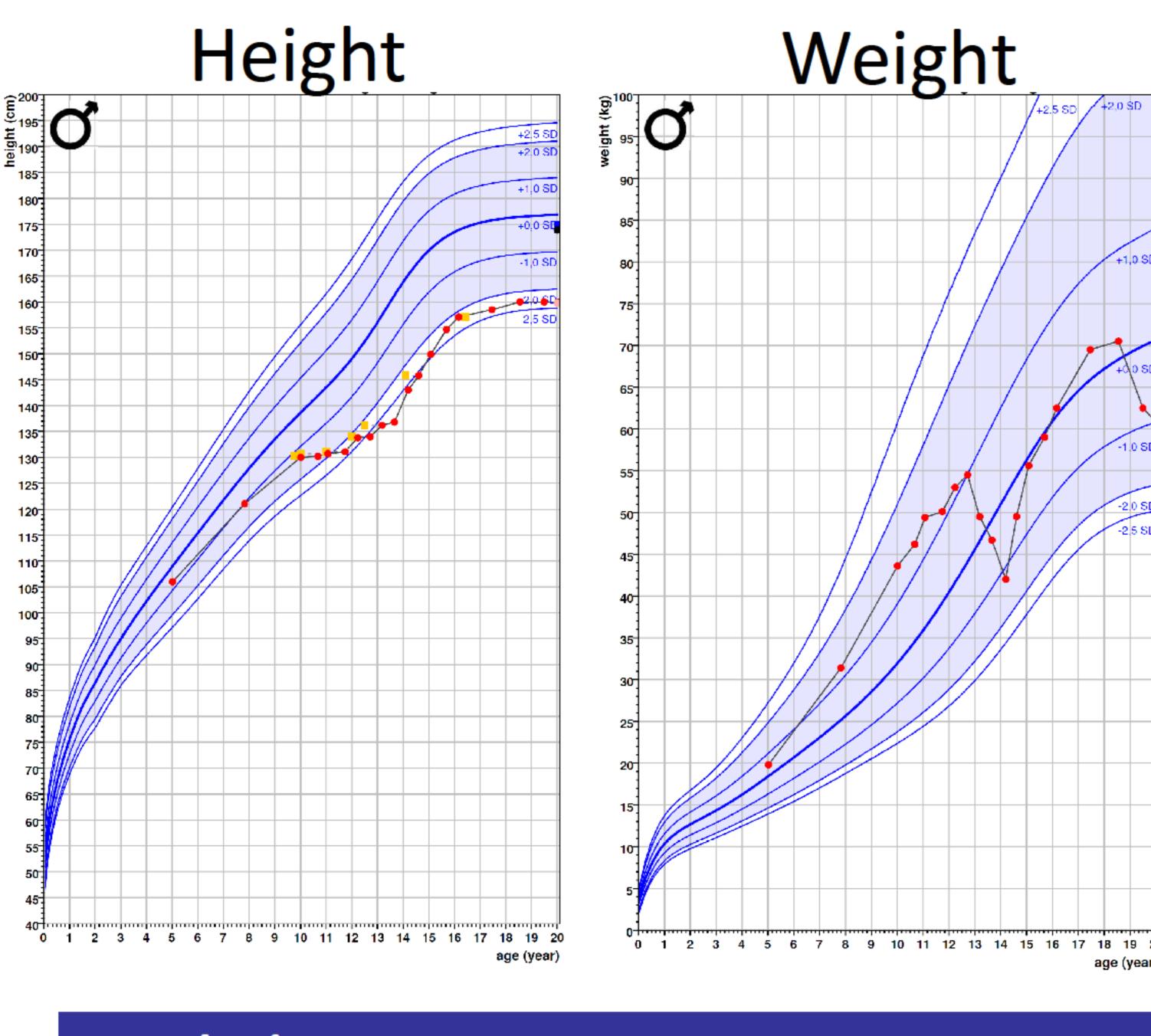
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## Background

First line treatment for an ACTH-secreting pituitary adenoma is transsphenoidal resection (TSR) of the tumor. Treatment options for patients with recurrent or persistent disease after surgery include another TSR, medical management, bilateral adrenalectomy, radiosurgery, or a combination of these. Radiosurgery represents a potentially curative treatment option for patients with recurrent or persistent disease.

## Case report

A boy was diagnosed at 10 years of age with an ACTH-secreting pituitary adenoma on the basis of symptoms (poor growth velocity, excessive weight gain and headache), laboratory findings (alterations of cortisol and ACTH circadian rhythm, cortisol and ACTH levels unsuppressed by dexamethasone suppression test, elevated urinary cortisol levels) and MRI imaging (4 mm microadenoma). He underwent TSR, but after few months ACTH and cortisol levels remained persistently high. As repeated neuroimaging were unable to identify the presence of a pituitary adenoma in spite of the persistent cushinoid features, we gave up the program to perform a new TSR and at the age of 13 years patient instead underwent a bilateral adrenalectomy followed by substitutive hormonal treatment with glucocorticoid and mineralcorticoid. After 18 months he developed a Nelson syndrome, characterized by very high ACTH levels and evidence of a microadenoma at the MRI. A second TSR was performed, but ACTH levels were persistently high after surgery. Then we decided to treat the patient with radiosurgery. At the age of 16 years he underwent a single treatment with CyberKnife (Dose 20 Gy). At the age of 22 years, after a 75 months follow-up period, patient is still in remission and does not exhibit other pituitary hormone deficiency.



	Cortisol (µg/dl)	ACTH (pg/ml)	Urinary cortisol (µg/24h)
At diagnosis (10 yrs 8/12)			488
h 8.00	17.9	39.6	
h16.00	24.1	39.2	
h 24.00	17.7	48.6	
After low-dose dexamethasone suppression test	16.2	35.2	
After high-dose dexamethasone suppression test	16.4	40	
Before arenalectomy (13 yrs 4/12)			355
h 8.00	18.6	50.1	
h16.00	15.5	85.9	
h 24.00	16.9	91	
Before 2nd TSR (14 yrs 9/12)			
h 8.00		>1250	
Before radiosurgery (15 yrs 11/12)			
h 8.00		>1250	

## Conclusions

Radiosurgery is a potentially curative treatment option in patients with recurrent and persistent pituitary adenoma. CyberKnife is a relatively new technology, but represents a valuable alternative option to the well-known Gamma Knife, as demonstrated in our patient.

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