

Capecitabine and temozolomide (CAPTEM) treatment of atypical macrocorticotropinoma in a patient with Nelson's syndrome

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Introduction. Atypical pituitary adenomas are often characterized by rapid growth and massive invasion of the surrounding structures. Usually, they are associated with poor prognosis and high recurrence rate due to resistance to conventional therapies. When surgery and radiation are ineffective, alternative therapies remain the last line treatment. Lately, a novel chemotherapy with capecitabine and temozolomide [1,2] was proposed as a highly effective and extending the life of the patient procedure in refractory corticotroph pituitary tumors.

Aim of study was to present an effectiveness of CAPTEM in atypical corticotropinoma in a patient with Nelson's syndrome.

Case report. 56-year-old male with Cushing's disease established 10 years earlier. During the first MRI 10x9x8mm pituitary adenoma was detected. Initially the patient underwent 3 subtotal selective transsphenoidal adenomectomies (2005, 2007, 2009). A postoperative pathologic exploration revealed a densely granulated corticotroph cell adenoma with MIB-1 index>10% and MGMT (-). Due to a re-growth of the tumor, the patient underwent consecutive stereotactic radiotherapy (9Gy) and received cabergoline for 19 months. Afterwards, because of intractable complications of hypercortisolism, as well as previous treatment failures, a total bilateral adrenalectomy was performed as a lifesaving procedure. Subsequently, the patient developed Nelson's syndrome with intense skin hyperpigmentation and aggressive pituitary tumor progression. In 2011 transcranial neurosurgery and tomotherapy (45Gy) were applied. Due to next re-growth of the tumor with its expansion to cavernous sinuses and suprasellar region with a compression of the optic chiasm, and exhaustion of all conventional therapeutic options, the patient was admitted to our department for qualification to Temozolomide [TMZ]. 2 cycles of TMZ [150 mg/m²] for 5 days were applied and because of the progression of eye damage CAPTEM was introduced. After 4 cycles hormonal, clinical and imaging stabilizations are observed.

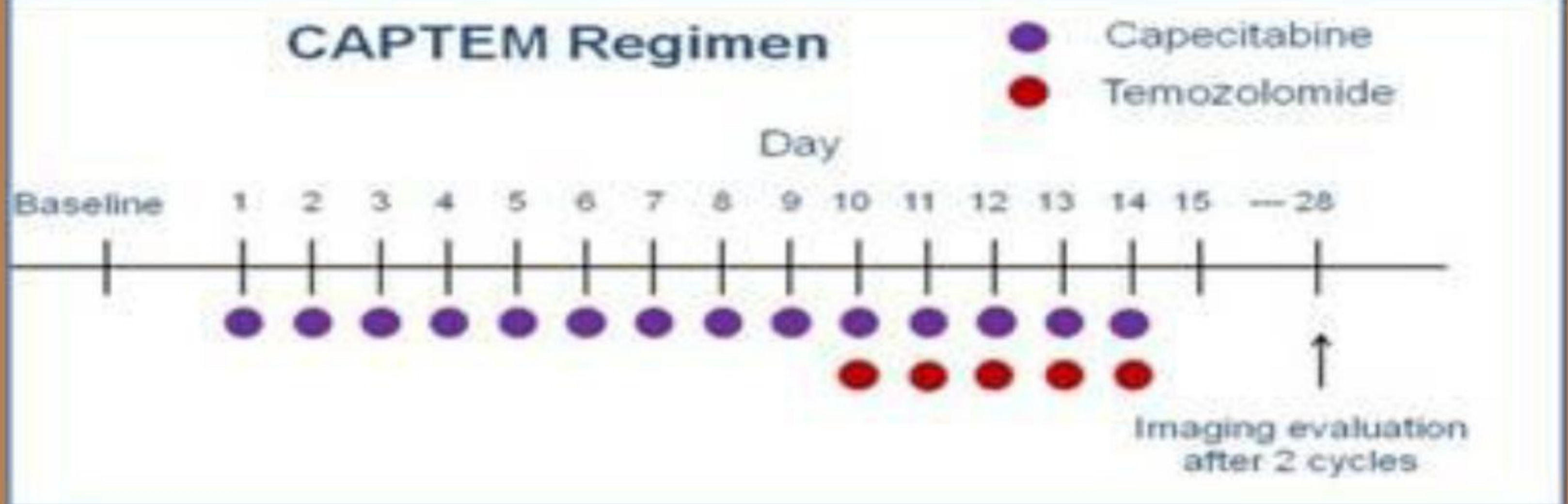
Conclusion. CAPTEM can be an effective treatment option in atypical adenomas in Nelson's syndrome.

References

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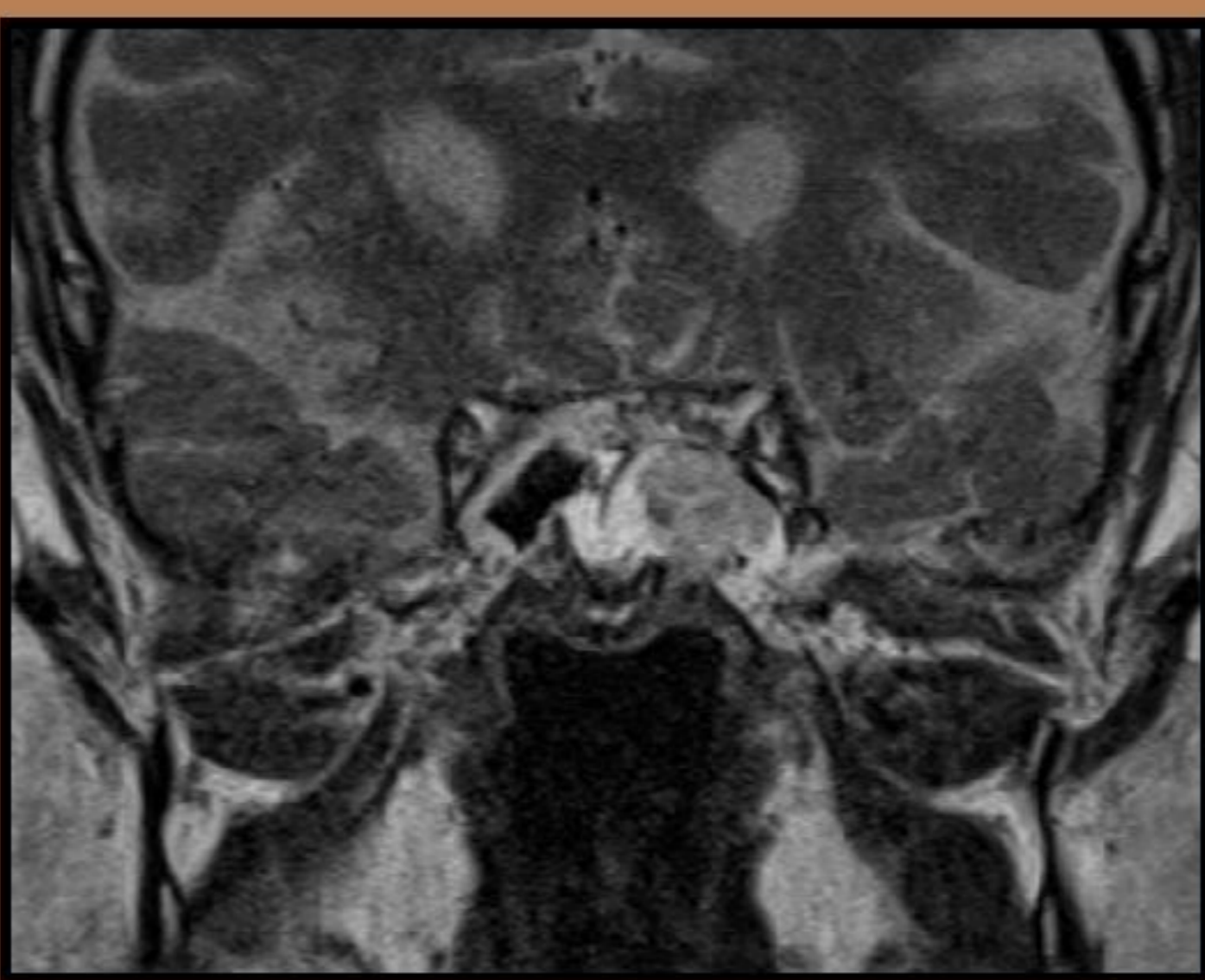
Schema of CAPTEM regimen. [JOP. J Pancreas (Online) 2013 Sep 10; 14(5):498-501].

Capecitabine in dose of 1,000 mg orally, twice daily on days 1-14 and temozolomide 200 mg/m² in two divided daily doses, on days 10-14 of a 28-day cycle



Tab.1. Disease control before and after CAPTEM treatment

	Before TMZ therapy	After 2 cycles of TMZ	After 4 cycles of CAPTEM
ACTH n. 7,2- 63,6pg/ml	1878	2640	1443
Tumor's size in MRI [mm]	12x14x12	35x26x25	32x17x21



Pituitary macroadenoma located in sella turcica and suprasellar region, penetrating to both cavernous sinuses and surrounding the optic chiasm