Association of two aggressive tumors: prolactinoma and multiple meningioma – difficult issue, difficult management

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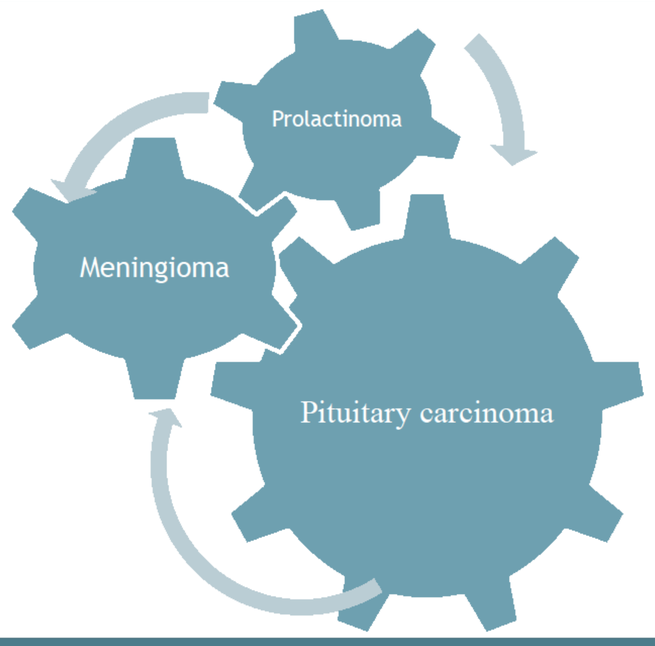
Introduction

❑ Pituitary adenomas are common intracranial tumors mainly considered as benign.

❑ Rarely, these tumors can exhibit an aggressive behavior, invading surrounding tissues, presenting a resistance to conventional treatment leading to early and frequent recurrences.

❑ Even more rarely, pituitary tumors can give rise to cerebrospinal or systemic metastases, therefore being qualified as pituitary carcinomas.

❑ Pituitary carcinomas are exceedingly rare, with an incidence of 0.2% of symptomatic pituitary tumors [1].



❑ Meningioma are benign tumors that derive from arahnoid membrane, with higher frequency in females than males [2].

❑ The coexistence of pituitary adenoma and meningioma is very rare.

❑ The association between prolactinoma and meningioma is partly attributed to the existence of prolactin receptors at the level of meningioma [3]

Case report

2008
46 year old female patient with bitemporal hemianopsia
no other clinical complaints

MRI: pituitary macroadenoma (22/19/35mm) with suprasellar evolution

hormonal balance

•hyperprolactinemia
PRL= 66ng/dl - X 3N (N: 1.2-19 ng/dl)

=> **Cabergoline** with *good evolution*

• secondary thyroid and gonadal insufficiency
TSH= 0,4 uUI/ml (N: 0.4-4uUI/ml)
FSH= 7.3uUI/ml (N: 35- 151uUI/ml)
LH = 3.5uUI/ml (N: 16-90uUI/ml)

3 years later 2011

• acute intracranial hypertension

⇩

Transcranial adenomectomy

• Gamma knife radiation

⇩

subsequent adrenal insufficiency

•Nodular goiter

⇩

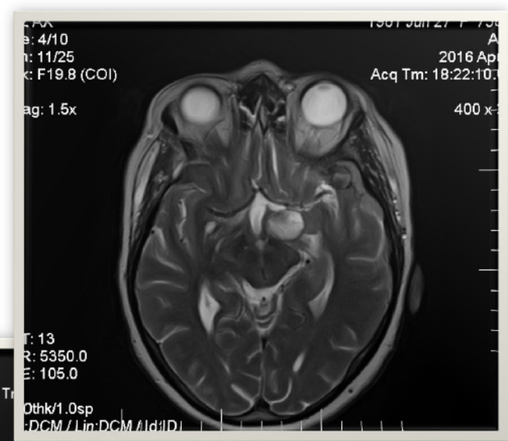
partial thyroidectomy

4 years later 2015

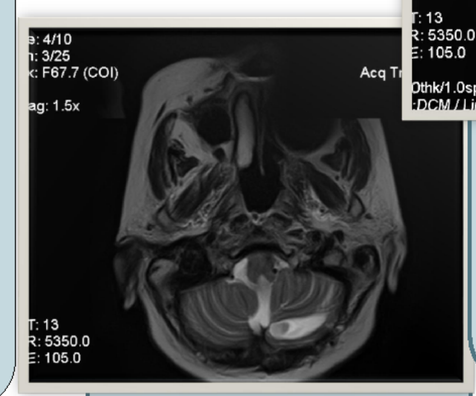
progressive tumor growth

⇩

second transcranian adenomectomy



Tumoral lesion with mixt structure: parenchymatous and chystic intra and extra sellar with extension in left cavernous sinus



MRI: nodular lesion with the aspect of meningioma Ponto-cerebral angle

1 year later 2016

• intensive vertiginous syndrome
• second episode of acute intracranial hypertension

MRI: two cerebral meningiomas

One located at craniospinal junction
=>urgent excision

Hysto pathological:

Disseminated eosinophilic pituitary adenoma

Post op: Left cranial nerves paresis X, XI, XII

Remnant:
aggressive pituitary adenoma vs meningioma

2016

Hormone	Values	Normal Values
TSH	0.221 uUI/ml	0.4-4uUI/ml
fT4 (under subtitution)	0.880ng/dl	0.89-1.76ng/dl
Cortisol (under substitution)	2.97 ug/dl	5-25ug/dl
IGF	64 ng/ml	87-238ng/ml
FSH	0.67mIU/ml	21.7-153mIU/ml
PRL	15.1ng/ml	1.9-25ng/ml

⇩ Pituitary carcinoma?

Meningioma ? ⇧

immunohystochemistry

negative: Anti S100, anti CK, anti EMA
low positive: anti NSE, anti Vim
KI 67= 2%

Discussions

❑ Pituitary adenoma express different receptors for Fibroblast growth factor FGF 1 and FGF2 . There is a high immunoreactivity towards circulatory FGF like in patients with sporadic pituitary adenoma and meningioma [3,4]. This fact could explain the association between adenoma and meningioma

❑ It is debatable if meningioma result as a consequence of hormone dependent growth or secondary to radiation, characterized by younger age at presentation, higher male-to-female ratio and biologically more aggressivenes compared to primary spontaneous meningioma [5].

Conclusion

❑ Patient's evolution is marked by rapid and extensive tumor progression in spite of adequate treatment, consistent with an aggressive pituitary adenoma.

❑ Nevertheless, in our case we discuss about the association between prolactinoma and multiple meningioma versus aggressive pituitary tumor.

❑ Further histopathogical and molecular markers could be helpful in establishing a firm diagnosis and targeted treatment.

References:
[1] Chatzellis E., Alexandraki K., Androulakis I., Kaltsas G., Aggressive pituitary tumors, Neuroendocrinology 2015;101:87-104 [2]Barnholtz-Sloan J., Kruchko C, Neurosurg Focus 23 (4):E2, 2007;[3] Muccioli G, Faccani C, Lanotte M, Forni M and Ciccarelli E; Prolactin receptors in human meningiomas: characterization and biological role J Endocrinol 1997 Jun;153(3):365-71; [4]Amit A, Achawal S, Dorward N. Pituitary macro adenoma and vestibular schwannoma: a case report of dual intracranial pathologies. Br J Neurosurg 2008;22:695-6.1;[5]Ueba T, Takahashi JA, Fukumoto M, Ohta M, Ito N, Oda Y, *et al*. *Expression* of fibroblast growth factor receptor-1 in human glioma and meningioma tissues. Neurosurgery 1994;34:221-5; [6] Strojan P, Popovic M, Jereb B, Secondary intracranial meningiomas after high-dose cranial irradiation: report of five cases and review of the literature, Int J Radiat Oncol Biol Phys. 2000 Aug 1;48(1):65-73