

Case Report

Collision tumor of meningioma and craniopharyngioma: a case report

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Abstract: Meningiomas are the commonest benign intracranial tumor, accounting for 13-26% of all primary intracranial tumors. They can arise from the dura at any site. Craniopharyngiomas are uncommon epithelial tumors which represent only 2-5% of all primary intracranial neoplasms and arise anywhere along the path of the craniopharyngeal duct. The co-existence of meningioma and craniopharyngioma has rarely been reported. Herein, we report an unusual case of an anterior fossa meningioma occurring together with a craniopharyngioma, which were radiologically not distinguishable in preoperative imaging. The clinical presentation, preoperative imaging, surgical treatment, histologic features of the two tumors and possible proposed mechanisms leading to the development of two different tumor types in one patient are discussed.

Keywords: Meningioma, craniopharyngioma, collision tumor, skull base

Introduction

Collision tumors are 2 separate growths which are developed simultaneously and in close proximity to each other [1-3]. Meningioma is the commonest intracranial tumor of the anterior and central skull base; craniopharyngioma is a relatively rare neoplasm which usually arises in the suprasellar region [4]. While the occurrence of these brain tumors in one patient which is well known in phakomatoses, such as in neurofibromatosis type 2 (NF-2), or with a history of previous irradiation of the brain [5, 6]. It is a very rare case without these circumstances. To our knowledge, this is the third one of this type of collision tumor to be reported.

Case report

History

A 57-year-old man with no history of neurocutaneous disorders and prior radiation therapy was referred to the department of Neurosurgery with a 1-year history of progressive both sides visual loss and intermittent headache. After a complete ophthalmologic evaluation, he performed an eye campimetry that showed severe decrease of visual acuity associated with bitemporal hemianopsia. The remainder of

the neurological examination proved to be normal. His endocrinological assessment was Testosterone 3.21 ng/ml [4.30-25.56], Cortisol (h:8.00) 31.40 ng/ml [50-250], FSH 6.56 mIU/ml [0.7-11.1], LH 1.62 mIU/ml [0.8-7.6], HGH 0.231 ng/ml [0-3], ft3 3.65 pmol/L [2.63-5.70], ft4 12.96 pmol/L [9.00-19.04], TSH 1.234 ulu/ml [0.350-4.940]. Afterwards, CT and MRI imaging of his brain were performed (**Figure 1**).

Neuroradiology

A computed tomographic scan showed a slightly hyperdense lesion in the anterior fossa and no calcifications within the tumor. MRI of the brain showed a skull base tumor which was composed of two parts. The anterior part is isointense on T1- and T2-weighted sequences and presents a dural sign. The posterior portion of the lesion is isointense on T1-w and hypointense on T2-w sequences, the solid component enhancing obviously after gadolinium administration.

Intervention

In order to save his eyesight, removing the tumor and decompressing optic pathways is in

A case of anterior fossa meningioma

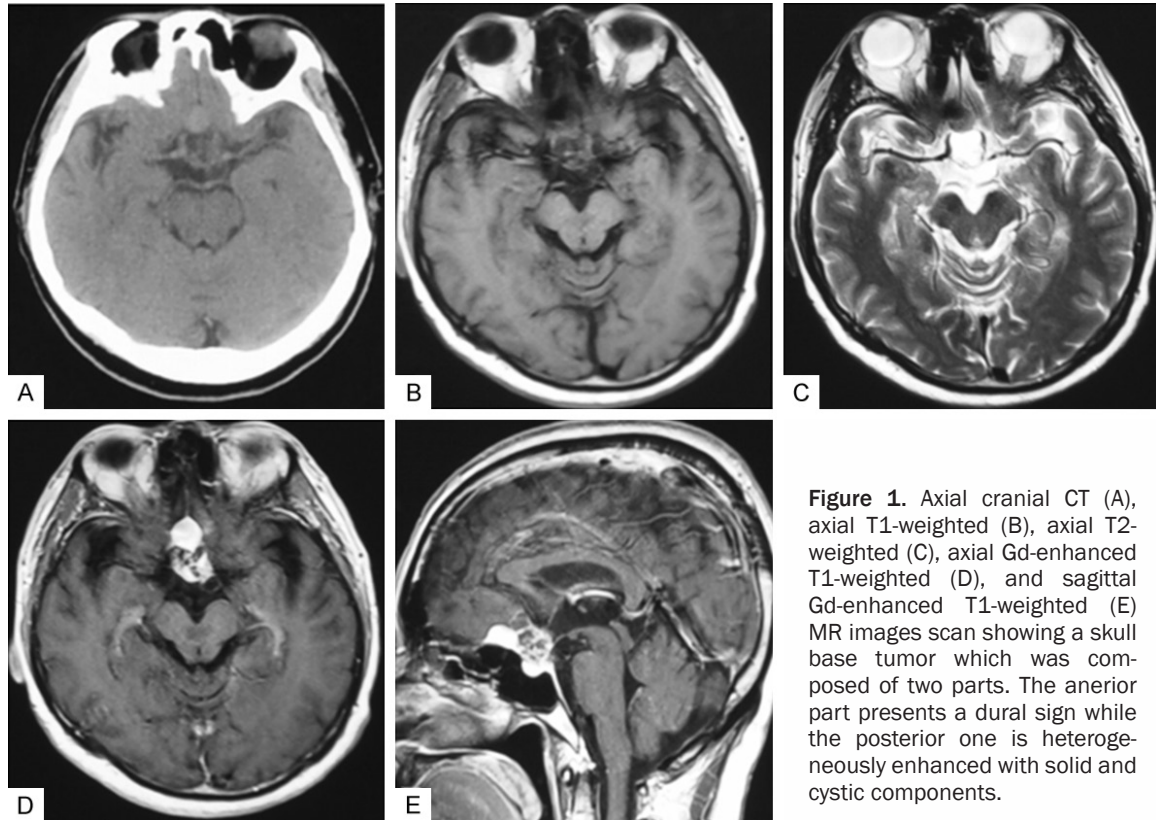


Figure 1. Axial cranial CT (A), axial T1-weighted (B), axial T2-weighted (C), axial Gd-enhanced T1-weighted (D), and sagittal Gd-enhanced T1-weighted (E) MR images scan showing a skull base tumor which was composed of two parts. The anterior part presents a dural sign while the posterior one is heterogeneously enhanced with solid and cystic components.

highly needed. He was brought to the operating room and positioned in horizontal position. A fronto-temporal craniotomy on the right side was performed, the sylvian cistern was opened, the frontal lobe retracted and finally the tumor exposed under the operating microscope. We surprisingly found that the mass was composed of two separate tumors, a craniopharyngioma and a meningioma with clear broad attachment to the dura of the anterior cranial base. Both tumors were completely removed and the dural base was extensively coagulated.

Pathology and postoperative course

Histopathological workup of both tumor samples confirmed the diagnosis of angiomatous meningioma and adamantinomatous craniopharyngioma (grade I sec. WHO classification) (**Figure 2**), respectively. The postoperative course was uneventful, except for a slight decrease of his visual condition was noted. At the most recent follow up (35 months after surgery), he was symptom free with no radiological recurrence.

Discussion

The simultaneous occurrence of meningioma and other type's intracranial tumors in the same patient has often been reported in literatures since meningioma is the commonest intracranial tumor of the skull base. Patients with meningioma and craniopharyngioma are rare, to date, only five cases have been reported in the English literature, including our patient, their clinical features are summarized in **Table 1** [7-10]. **Table 1** summarized 5 cases of tumors found in two men and three women with ages ranging from 54 to 81. Among the previous cases only two patients with the two tumors in contiguity (case 3, 4). In the two cases, only tumors in case 4 were originated from the skull base, since the location of the craniopharyngioma in case 3 were in the third ventricle [9, 10]. However, among the 4 previously described cases, 3 occurred in the Italian population (case 1, 2, 4) and 1 occurred in the USA (case 3). The present case was the first to occur in a Chinese patient and consequently widening the epidemiological spectrum of this type collision tumor.

A case of anterior fossa meningioma

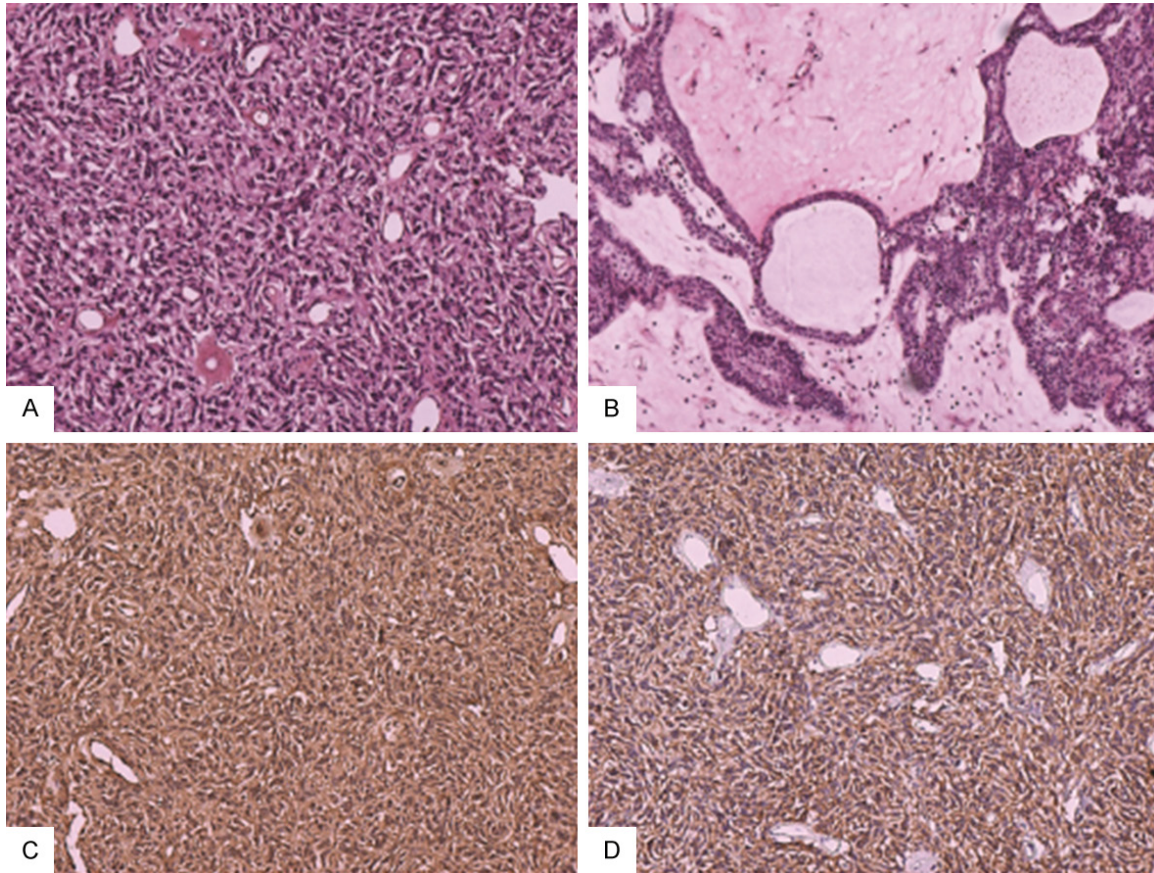


Figure 2. Histological images. (A) Angiomatous meningioma characterized by foci of meningothelial cells and highly vascular structures (H&E, original magnification $\times 100$); (B) Adamantinomatous craniopharyngioma is recognized by the presence of multi stratified squamous epithelium and peripheral palisading of columnar cells (H&E, original magnification $\times 100$); (C, D) Immunohistochemical studies showing a clearly positive reaction for vimentin (C) and epithelial membrane antigen (D) in the meningioma.

The incidence of meningioma is highest after the fifth decade of life while craniopharyngioma has two peaks of incidence, from 5 to 10 years and 50 to 60 years of age [11, 12]. In the presented case, the patient's age was coincident with the epidemiology of the two tumors.

Meningiomas are well-defined, extra-axial masses, they are most commonly isointense or slightly hypointense to brain on T1-weighted imaging and hypointense on T2-weighted imaging, after gadolinium administration, meningiomas show strong homogeneous enhancement. Most commonly, craniopharyngiomas arise in the suprasellar/parasellar region. It is a largely cystic or mixed tumor, with calcifications and solid component enhancing after gadolinium administration. Depend on the protein content of the fluid the cystic component may have variable T1-weighted signal intensity while it is usu-

ally hyperintense on T2-weighted sequences [13, 14]. The term 'collision' has been used for the cases in which two tumors are being intermixed or appear close to each other without brain tissue in between [15]. Due to the special location relationship of the two tumors, the pre-operation diagnosis of two separate tumors was challenging, though a correct radiological diagnosis is critical to plan the treatment strategy. So a differential diagnosis of co-localization tumors should be kept in mind while dealing with such pathology.

Patients with phakomatoses, such as NF-2 and a previous history of irradiation are well-known causes for the development of multiple primary brain tumors [16-19]. As for our patient, he had not received prior radiation, moreover he had no family history and no clinical signs of phakomatoses, although gene detection for NF-2 was

A case of anterior fossa meningioma

Table 1. Literature review for the coexistence of meningioma with craniopharyngioma

Case	Report year	Age, sex	Symptom, duration	Meningioma	Craniopharyngioma	Treatment	Post-operative course	Follow up
1	1967	54, female	Serious behavior disorders associated with an evident "change of character", 7-8 months	A left frontal parasagittal meningioma	Invading the third ventricle	-	-	Died suddenly
2	1981	65, male	Early morning midfrontal headaches and intellectual deterioration, 3 months	A small meningioma en plaque between the anterior clinoids	In the anterior 3 rd ventricle extending into the hypothalamus	Transfrontal craniotomy	NA	Alive and well
3	2005	61, female	Headache and bilateral visual loss, 3-month	Tuberculum sellae meningioma (about 2 cm in diameter)	Occupied the third ventricle (3.5 cm)	Right pterional approach. Complete removal of the tuberculum sellae meningioma and subtotal excision of the craniopharyngioma.	Died ten days postoperatively for diencephalic failure.	-
4	2011	81, female	Increasing left eye visual loss, bitemporal hemianopsia and bifrontal headache, 1 month	Originating from the anterior cranial base dural surface	Arising from the anterior superior margin of the pituitary gland	Right fronto-temporal craniotomy, both tumors were gross totally removed.	Good recovery, eyesight modest improvement	Alive and well
Present case	2012	57, male	Progressive both sides visual loss and intermittent headache, 1 year	Anterior cranial base	Suprasellar	Right fronto-temporal craniotomy, both tumor were completely removed.	Uneventful	Alive and well

A case of anterior fossa meningioma

not performed on him. Several hypotheses have been proposed to explain the coincidence of two completely separate primary brain tumors of different histogenesis in the same patient: (1) tumors can develop entirely coincidentally; (2) the initial tumor can act as a stimulus on the surrounding cerebral parenchyma or meningeal tissue to induce a new tumor in different tissue; (3) a carcinogenic stimulus may develop tumors in different tissues simultaneously; or (4) a residual embryonic structure becomes the basis for subsequent multiple brain tumor development. Furthermore, several studies have addressed the role of endogenous as well as exogenous hormonal therapy on the risk of meningioma formation [20]. Craniopharyngioma patients present endocrine disturbances in 80-90% [21]. So, we suggest that the hormonal status maybe another cause for the coincidence of meningioma and craniopharyngioma. More research on possible mechanisms leading to the simultaneous occurrence of meningioma and craniopharyngioma has to be done to support this theory.

In conclusion, we have reported what is, to our knowledge, the third one of this type of collision tumor arising in a Chinese patient. The concurrence of meningioma and craniopharyngioma is rare, especially is this type of collision tumors which represents a serious difficulty in imaging and clinical diagnoses. When skull base tumors with imaging examination uncharacteristic are detected, particularly in the elderly patients (since the age of the patients mentioned in **Table 1** are all above 50), the diagnosis of colocalization tumors should be kept in mind. More cases of this type collision tumor must be studied to draw definitive conclusions about its mechanism.

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Disclosure of conflict of interest

None.

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A case of anterior fossa meningioma

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