Giant tuberculum sellae meningioma with unruptured anterior communicating artery aneurysm encased
Case report and review of the literature

Bogdan Constantin Dumitrescu¹, Mircea Radu Gorgan²

¹PhD Student in Neurosurgery, “Carol Davila” UMPh Bucharest
Faculty of Medicine, Department of Neurosurgery
²Clinic of Neurosurgery, Emergency Clinical Hospital “Bagdasar-Arseni”, Bucharest

Abstract
This 64-year-old man presented with symptoms of optic chiasm and nerve compression from a giant tuberculum sellae meningioma with unruptured anterior communicating artery aneurysm encased. Preoperative magnetic resonance imaging disclosed that the aneurysm was completely enclosed in the tumor, but angiographic studies did not reveal any important arterial narrowing. The embedded aneurysm caused neither SAH nor intratumoral hemorrhage. All of these factors pointed to little adhesion between the tumor and the encased arteries. Surgery was performed a week after admission and the intraoperative findings revealed that the tumor did not adhere too much to the enclosed vasculature except for a little part attached to the left ICA. The aneurysm was safely clipped after piecemeal removal of the tumor, which was finally extirpated without fear of aneurysm rupture, and with a good visual outcome. To treat both the tumor and aneurysm simultaneously, careful stepwise operative procedures were essential.

Keywords: aneurysm, anterior communicating artery, meningioma, tuberculum sellae

Introduction
Meningiomas account for about 30% of all primary brain tumors diagnosed in adults, and has an estimated incidence in women approximately twice those in men. The vast majority of meningiomas are considered histologically benign (over 90%) (5). Meningiomas of the tuberculum sellae comprise approximately of 3 to 10% of all intracranial meningiomas and they arise from the limbus sphenoidal, chiasmatic sulcus, and tuberculum sellae. For a correct diagnosis and management it is required an appreciation of the unique clinical, neuroimaging, and surgery-related features that distinguish these meningiomas from others of the anterior skull base.(6, 20) Tuberculum sellae meningiomas characteristically use to lie in a suprasellar subchiasmal midline position, displacing the optic chiasm posteriorly and slightly superiorly, and the optic nerves laterally. The most common initial complaint is the slowly progressing visual deterioration, and prompt treatment is intended to preserve and improve vision. Ideally management consists of gross-total resection without injury to neighboring vital structures.(6) Various approaches to the tuberculum sellae region are used for resection.
There are a variety of craniotomies (pterional, unilateral subfrontal, bilateral subfrontal, and so forth) that are being used in an attempt to find the most direct route to this region with the fewest potential complications or anatomical limitations.

An association of brain tumor and intracranial aneurysm rarely occurs. Aneurysms have been observed in patients with gliomas, pituitary adenomas, and other rare brain tumors, but the coincidence is highest in patients with meningiomas. (4, 12, 21) Although a number of reports of meningiomas coexistent with intracranial aneurysms have been published, there are not many cases of meningiomas with encased unruptured aneurysms. (3, 4, 7, 12, 14, 16, 19, 21, 22)

We report a case of an elderly patient with unruptured anterior communicating artery aneurysm completely encased in a giant meningioma of the tuberculum sellae, treated into the Forth Department of Neurosurgery. We reviewed medical records, imaging, treatment and follow-up.

Case report

A 64-year-old man presented at the neurosurgery department in our hospital with progressive onset of a left eye blindness, decreased right eye visual acuity, anisocoria (left pupil larger than right), diffuse headache, dizziness and balance disorders. He experienced no disturbance of consciousness, other cranial nerve abnormalities, motor paresis, or sensory impairment. The patient had a long history of hypertension, but no other pathology was noteworthy.

Examination. On admission, the patient was neurologically intact except for left eye blindness, decreased right eye visual acuity and anisocoria.

Gadolinium-enhanced T1-weighted MR images disclosed a giant extraaxial suprasellar high-intensity lesion with left parasellar development, measuring 44/41/48 mm, with homogeneous contrast enhancement, which was isointense on T1-weighted imaging; these images also suggested that the aneurysm was completely encased in the tumor (Figure 1). Angio-RM and four vessels cerebral DSA demonstrated an aneurysm arising from the ACoA and a faint patchy stain around the sella and parasellar on the left (Figure 2 and 3). The bilateral ICAs and ACAs were shifted posteriorly, ascending and to the right, but no arterial narrowing was observed (Figure 4). The tumor is fed by the left middle meningeal artery from the external carotid artery and both ICAs. The aneurysm is supplied by the right ICA and has a dome of 5.22 mm in diameter and a neck length of 3.38 mm. Pulmonary X-ray and electrocardiography were normal, except for aortic atheromatosis.

The operation was performed on the 7th day after admission. The patient received Dexamethasone 16 mg/d, Furosemide 40 mg/d, antiepileptic drugs, analgesics and intravenous hydration. After the patient received a general anesthetic, he is positioned supine on the operating room table, with the neck flexed on the chest and the head extended on the neck. A standard bicornoral skin incision was made followed by a bilateral frontal craniotomy with frontal sinus opening and cranialisation. Dural opening was parallel with the skull base with frontobasal ligation and cutting of the superior sagittal sinus. Via the interhemispheric approach, we identified pericallosal arteries and genu corpus callosum.
Figure 1 Cerebral MRI, giant frontobasal extraaxial tumor, measuring 44/41/48 mm, with homogeneous gadolinium enhancement, with dural insertion at tuberculum sellae, and suprasellar and left parasellar extension.
The pericallosal arteries were followed back towards the ACoA and a gray-reddish, relatively firm tumor was encountered. The tumor displaced the optic chiasm and bilateral ACAs superiorly and posteriorly.

We identified the right optic nerve, right ACA and ACoA containing the aneurysm is found encased in the tumor. We performed piecemeal resection, dissected the right
ACA and ACoA. The aneurysm, which originated from the right part of ACoA, was dissected, encountering no adhesion between the tumor and the neck of the aneurysm. A right 11 mm Yasargil vascular clip was used to obliterate the unruptured aneurysm successfully. Without fear of premature rupture, the residual tumor on the dome and above the sella turcica was resected after clipping. The tumor was totally removed, except for a little part attached to the left ICA, extended in the left cavernous sinus. The attachment at the tuberculum sellae was entirely coagulated (Simpson Grade 4). We performed wound closure with watertight duroplasty with pericranium and anchoring of dura mater, epidural drain, replacement and fixation of the bone flap and skin suture. The pathological diagnosis was meningotheliomatous meningioma. No hemosiderin deposit was found anywhere in the specimen.

The patient’s postoperative course was uneventful except for bilateral anosmia. His left eye visual acuity considerably increased, but the right eye remained blind. The patient was discharged in good condition on the 9th day after surgery.

Discussion

Estimated incidence of aneurysms in patients with brain tumors is reported as 0.2 to 0.7%, which is most likely lower than the true incidence because four-vessel angiographic studies are not always performed in patients harboring intracranial tumors. Scamoni et al., reported that the meningioma and aneurysm were observed on the same side in more than 80% of the patients reported in literature. Sometimes, when subarachnoid hemorrhage occurs, it is always secondary to aneurysm ruptures.

The case we report, as far as we know, is one of the few anterior communicating artery aneurysms encased in a tuberculum sellae meningioma.

Several mechanisms of aneurysm formation associated with meningiomas have been proposed. Kandel, et al., described a patient in whom a saccular aneurysm of the middle cerebral artery was enclosed in a frontotemporal meningioma. Under microscopic examination, the tumor was found to be intimately attached to the arterial adventitia. These authors ascribed development of the aneurysm to the damage to the middle cerebral artery caused by the meningioma. Although we did not examine the aneurysm wall microscopically, in our case we presumed that the tumor had invaded at least partially the arterial walls but not necessarily the aneurysm, because the lesion was relatively easily dissected from the aneurysm and the enclosed vasculature, except for a little part of the tumor attached to the left ICA. Pia, et al., attributed the mechanism to high regional blood flow to the tumor. The tumor displaced the arterial structures leading to vascular distortion and kinking, with consequent blood flow change. In our case the meningioma is a highly vascularized lesion, being fed by middle meningeal artery from left external carotid artery and bilateral ICAs. We also considered other factors such as stimulated vascular proliferation, a particular genetic
background or simple coincidence, because the aneurysm did not arise on the main feeding vessel to the tumor. (8, 13)

Treating the aneurysm is mandatory because otherwise rupture risk is important. The treatment options are a two staged therapy, the first being embolization of the aneurysm and arterial feeders of the tumor followed by surgical removal of the meningioma. Another option is only surgical treatment resolving both lesions in the same operation. We chose the surgical only solution because our patient already had left eye blindness and decreased right eye visual acuity, and we did not want to further delay the treatment. The bilateral frontal craniotomy was chosen because it produces a wide surgical view between the two ICAs, allowing the lesion to be approached via various routes. Otherwise an ordinary pterional or unilateral frontal craniotomy would have been enough to avoid damaging the olfactory nerves, but in our patient the tumor was not soft enough and partially adhered to the encased vasculature, especially to the left ICA. (2)

Unfortunately we cannot determine preoperatively whether it will be possible to easily dissect the tumor from the encased vasculature. Sekhar and Javed stated that the presence of an arachnoidal plane is inferred during surgery by the ease of dissection. (18) Kawase, et al., emphasized the difficulty of dissecting meningiomas from the encased arteries without the presence of the arachnoidal plane. (11) Al-Mefty classified clinoidal meningiomas into three categories according to the presence of arachnoid membranes between the tumor and cerebral vessels. In his Group 1, the tumor adheres directly to the adventitia, which prevents total removal and leads to poor outcomes. Preoperative angiographic studies demonstrate narrowing of the encased arteries in this group. (1) In our case, however, the embedded aneurysm caused neither SAH nor intratumoral hemorrhage, and preoperative angiographic studies did not reveal important arterial narrowing. All these factors combined might have indicated that a regional pial or arachnoidal layer was kept relatively intact, which led to a limited adhesion between the tumor and vascular structures except for a small part of the left ICA. (1)

Preservation of vision is the most important goal of treatment and we agree that the key to success in preserving vision is adhering to arachnoid planes around the optic nerves, chiasm, and brain parenchyma, as well as recognizing that the vascular supply to the optic apparatus is derived via perforating branches off the posterior ethmoidal, superior hypophysial, and ACAs. (6) Careful stepwise procedures are essential to treat the aneurysm and the tumor simultaneously in a quite complicated case such as the one reported in this article. Because the aneurysm was incased in the tumor it was mandatory that we performed partial tumor resection to gain access to the vascular lesion. One of the technical challenges is the fact that right ICA supplied right posterior cerebral artery through the posterior communicating artery and also both ACAs, increasing the risk of cerebral ischemia in case of incidental injury. Another challenge is to identify the aneurysm in the tumoral mass, the main risk being intraoperative rupture. To do this, we exposed the pericallosal arteries, followed them backwards towards the ACoA. We started with piecemeal resection of the tumor to achieve precise anatomical orientation of the encased arteries. Then we carefully removed the
tumor around the aneurysm neck and applied a suitable 11 mm Yasargil vascular right clip. Another technical is challenge was the lack of proximal control on the parental vessel. The tumor acted like a seal to the aneurysm dome by canceling the transmural stress, preventing its rupture. After complete clipping of the aneurysm, we could then resect residual tumor on the dome without fear of rupture. Otherwise, clipping the residual tumor together with the aneurysm may lead to incomplete clipping or tumor regrowth. We performed a subtotal resection leaving a small piece of lesion attached to the left ICA, extended in the left cavernous sinus. For this residual tumor the patient should be referred for gamma-knife surgery. In cases like this one, a two-stage treatment is also possible, but the aneurysm could only be safely clipped after the aforementioned careful stepwise tumor removal. Embolization of the aneurysm in a first stage followed by late phase tumor removal is another alternative.

**Conclusions**

This is one of the few reports of an unruptured anterior communicating artery aneurysm completely encased in a tuberculum sellae meningioma. This type of tumors should be resected early in their course in patients presenting with symptoms of optic chiasm compression. Preoperative angiographic studies revealed encased arteries with almost normal diameters which generally indicated little adhesion between the tumor and the vascular structures. Although there are several surgical approaches to these tumors, we believe that the bifrontal approach yields minimal morbidity and allows the surgeon the greatest intraoperative flexibility for dissection and tumor removal. Using modern microsurgical techniques and a thorough knowledge of the microvascular anatomy of the blood supply to the optic nerves and chiasm, we successfully removed the tumor and clipped the aneurysm with an excellent visual good. Careful stepwise operative procedures were essential to treat both the tumor and the aneurysm simultaneously.

**Abreviations**

ACA – anterior cerebral artery  
ACoA - anterior communicating artery  
DSA - digital subtraction angiography  
ICA – internal carotid artery  
MR – magnetic resonance

**Correspondence and correction to**

Bogdan Constantin Dumitrescu, address: Emergency Clinical Hospital Bagdasar-Arseni, No. 10-12, Berceni Street, Sector 4, Bucharest;  
e-mail: dumitrescu.bog@gmail.com;  
tel. 0745.696.966

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