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Case Report

Spontaneous intracerebral pseudoaneurysm rupture and meningiomatosis: A case report and review of the literature

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ABSTRACT

Background: We report the first case of a spontaneous ruptured anterior cerebral artery pseudoaneurysm in a patient affected by meningiomatosis.

Case Description: A 71-year-old female patient was admitted to our emergency department after acute loss of consciousness. An urgent head CT scan showed third ventricle hemorrhage and a giant extra-axial tumor with associated peritumoral bleeding. A second, smaller, and right-sided tumor was detected at the posterior third of the superior sagittal sinus, indicative of meningiomatosis diagnosis. A following CT angiogram showed an hypervascularized lesion at the right frontal convexity and a ruptured A2 pseudoaneurysm. Tumor removal was performed through right frontal craniotomy. After the initial debulking and removal of the peritumoral hemorrhage, the A2 segment associated with the bleeding pseudoaneurysm was surgically coagulated.

Conclusion: We report the unique occurrence of two relatively rare neurological entities: meningiomatosis and intracranial pseudoaneurysm. In our experience, their simultaneous and acute presentation is associated to poor prognosis.

Keywords: Giant meningioma, Intracranial hemorrhage, Meningiomatosis, Pseudoaneurysm

INTRODUCTION

Meningiomas are the most common primary intracranial tumors, representing 37% of all primary brain tumors. Meningiomatosis, however, is a rare neurological phenomenon accounting for <2% of all meningioma cases.[2] The occurrence of intercranial pseudoaneurysms is an even rarer event, with an estimated prevalence of <1% among of all intracranial aneurysms. We report the first case of a spontaneous ruptured anterior cerebral artery pseudoaneurysm in a patient affected by meningiomatosis.

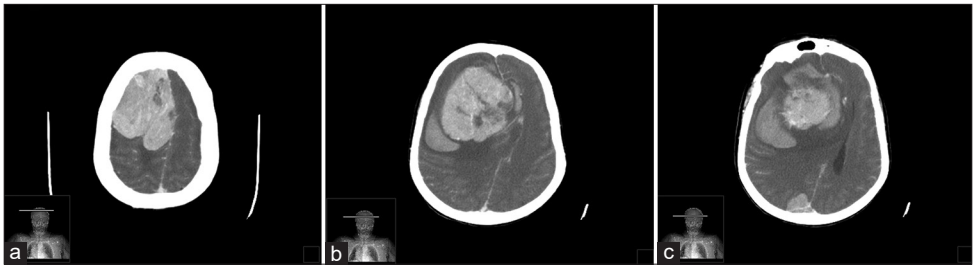

Figure 1: Axial CT with contrast: (a) right frontal convexity giant meningioma; (b) axial CT scan of the patient showing intracranial hemorrhage concurrent to the giant meningioma. Large peritumor brain edema and midline shift (2.5 cm) are also noted; (c) small 2 cm meningioma on the third posterior of the SSS.

CASE ILLUSTRATION

Clinical history

A 71-year-old female patient was admitted to our emergency department after acute loss of consciousness with a Glasgow Coma Scale score of 4/15. Neurological examination revealed anisocoric pupils (right>left), drowsiness, left-sided hemiplegia, and right-sided decerebration. An urgent head CT scan showed third ventricle hemorrhage and a giant extra-axial tumor with associated peritumoral bleeding [Figures 1a and b]. A second, smaller, and right-sided tumor (2 cm) was detected at the posterior third of the superior sagittal sinus, leading to a radiological diagnosis of cerebral meningiomatosis [Figure 1c].

A brain CT angiogram was performed, showing a hypervascularized lesion located at the right frontal convexity, with related mass effect and contralateral midline shift (2.5 cm). A ruptured A2 pseudoaneurysm (93.7 mm × 3.2 mm) was also documented [Figures 2a and b].

A right frontal craniotomy was performed. After the dura mater was opened, the tumor with the associated perilesional bleeding was exposed. The neoplasm was friable in consistence and was easily aspirable using and ultrasonic aspirator, showing macroscopic features concordant with a typical meningioma presentation. Although the macroscopic vascularization was prone to intraoperative bleeding, it was controlled with bipolar cauterization. After the initial debulking and removal of the peritumoral hemorrhage, the frontopolar artery was exposed through an interhemispheric dissection to reach the A2 segment associated with the bleeding pseudoaneurysm, which was then coagulated. We completed the procedure achieving a radical removal of the tumor and a satisfying hemostasis. Bone flap was replaced, and pathological tissue was sent for histological examination.

Histological examination

The histological analysis reported a fibrous meningioma (WHO Grade I) with diffuse intraparenchymal hemorrhage, without a brain parenchyma infiltration (EMA + and progesterone +).

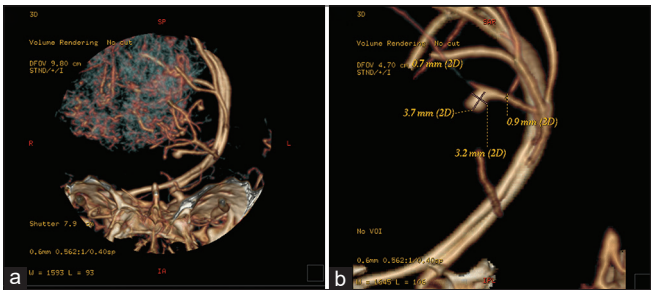

Figure 2: Anterior-coronal 3D CT angiogram: (a) vascularization of giant convexity meningioma; (b) A2 pseudoaneurysm (3.7 mm).

Postoperative course and postoperative imaging

After surgery, the patient was isochoric, and a further postoperative CT scan demonstrated common postoperative findings, including improvement of the midline shift and right frontal hypodensity related to the malacic brain parenchyma next to the meningioma [Figures 3a-c]. Unfortunately, the patient developed ab ingestis pneumonia, probably due to the sudden loss of consciousness, further worsened by mechanical ventilation, and died 5 weeks after surgery.

DISCUSSION

Meningiomatosis is a rare entity occurring in 1–2% of all meningiomas cases. It is defined as two or more meningiomas occurring simultaneously in different locations without an association with neurofibromatosis, as in the case of our patient. The pathophysiology of meningiomatosis is still not well understood. One theory suggests that the multiple tumors are a result of tumor cell migration through cerebrospinal fluid. In contrast, other studies have found evidence that points toward the independent evolution of the tumors.[4] The management of meningiomatosis primarily involves surveillance and observation. Surgical interventions are only considered when tumors begin to cause physical symptoms or show rapid growth. In the management of these patients, the tumor responsible for symptom onset is primarily targeted. However, simultaneous tumors that are

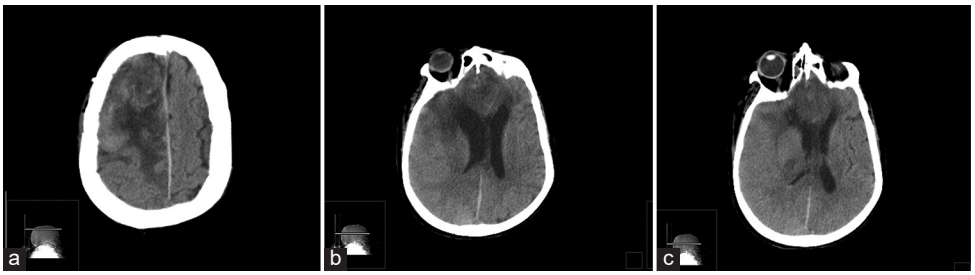

Figure 3: (a-c) Postoperative images show maximal meningioma debulking through ultrasonic aspiration, resolution of intracranial hemorrhage, and subsequent correction of midline shift.

readily accessible in the area are often also resected. The rate of postsurgery recurrence is around 13%, mostly in patients with high-grade meningiomas or that have undergone radiation therapy.[4] In general, Gamma Knife radiosurgery is the treatment of choice for small tumors in critical brain regions, such as eloquent cortex or areas adjacent to the visual pathways, for postsurgical remnants, or for treating small recurrences in elderly patients not eligible to undergo surgery. In addition, recent advancements in Gamma Knife instrumentation allow for implementation in larger tumors through hypofractionated sessions. Stereotactic radiosurgery is also showing promising results for treating multiple lesions.

The case we presented is the first association between pseudoaneurysms and meningiomatosis in an adult patient. The only other published case of pseudoaneurysms in association with meningiomatosis occurred in a pediatric patient.[3] Typical aneurysms involve all three layers of the artery, whereas pseudoaneurysms are characterized by the involvement of only the two outer layers, namely, the media and the adventitia.[1] Radiologically, this is indicated by irregularities of the aneurysm wall, delayed opacification of the lesion, and delayed washout and retention of the contrast material.

Individually, meningiomatosis and cerebral pseudoaneurysms have relatively straightforward management strategies with reasonable outcomes. However, as seen in the case we describe, the simultaneous presentation can necessitate rapid and complex surgical interventions, and yet still result in rapid clinical depression and dismal prognosis.

In the present case, the giant convexity meningioma may have contributed to the rupture of the adjacent A2 pseudoaneurysm. Indeed, the mass effect of the tumor caused increased intracranial pressure, responsible for a consequent rise in the intracranial blood pressure to maintain a constant brain perfusion. The process likely started a vicious cascade that leads to the bradycardia and irregular breathing seen in our patient (Cushing reflex). The rupture of the A2 pseudoaneurysm contributed to the intraventricular and peritumoral bleeding, further worsening the rise in the intracranial pressure and causing the sudden loss of consciousness and anisocoria that required urgent treatment.

Pseudoaneurysms are most frequently associated with trauma, but other etiologies include iatrogenic injuries, infectious disease, and connective tissue disease. The specific incidence of tumor-related pseudoaneurysms, such as the case we describe, is not known. The majority of intracranial pseudoaneurysms reported in the literature result from complications of tumor resection surgery.[5] Spontaneous occurrence has been reported after high-volume radiotherapy such as stereotactic radiosurgery, following radiation-induced damage of the external arterial layers. The most common treatment strategies for pseudoaneurysms include microsurgical approaches, such as clipping, wrapping, or ligation, and/or endovascular techniques, such as coiling, flow diversion, or parent artery occlusion. Endovascular surgery is less invasive and has become the modality of choice for low complexity cases. Microsurgery is a suitable alternative for situations in which endovascular treatment is not feasible, such as distal aneurysms or poor vascular morphology, or patients presenting with acute complications, such as the large hematoma seen in our patient.

CONCLUSION

We report the unique simultaneous occurrence of two relatively rare neurological entities in one patient, namely, cerebral meningiomatosis and intracranial pseudoaneurysm. Based on our experience, their simultaneous and acute presentation should be promptly managed but is still associated with poor prognosis.

Declaration of patient consent

Institutional Review Board (IRB) permission obtained for the study.

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Conflicts of interest

There are no conflicts of interest.

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