Cavernous angioma within an olfactory groove meningioma

Case report

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The authors present the case of a 60-year-old woman who was admitted to their institution after suffering a subarachnoid hemorrhage (SAH). Neuroimaging data demonstrated an olfactory groove meningioma surrounded by a slight edema, but there was no evidence of SAH, although results of the lumbar puncture demonstrated xanthochromic cerebrospinal fluid. Angiography confirmed the diagnosis of meningioma, but results of magnetic resonance imaging led the authors to suspect a cavernoma within the meningioma. This diagnosis was established by pathological examination of the resected lesion. The patient did well and was discharged soon after surgery. This very rare association and the propensity of each of these lesions to be revealed by hemorrhage are discussed.

Key Words • cavernous angioma • meningioma • brain neoplasm • olfactory tract • skull base

Skull base meningiomas and cerebral cavernous angiomas are quite frequently encountered in a neurosurgical practice. The association between these two entities is nevertheless very uncommon and, as far as we know, has never before been reported in the literature. We present a case in which this unusual association was found, and we discuss the propensity of each of these lesions to be revealed by intracranial hemorrhage.

Case Report

History, Examination, and Neuroimaging Findings. This 60-year-old woman presented to the emergency department with unusual, recent, and severe headaches, a fever of 38°C, and vomiting. The patient’s neurological examination revealed no other abnormalities. A CT scan obtained with and without a contrast agent revealed an olfactory groove meningioma surrounded by a slight edema (Fig. 1 left). There was no apparent SAH. To eliminate definitively the presence of this condition, a lumbar puncture was performed, and this test demonstrated the presence of an SAH. The cerebrospinal fluid contained 200 cells/mm³ (60% polymuclear; 40% lymphocytes); numerous red blood cells; a glucose level of 0.55 g/L; a protein level of 2.18 g/L; and it was xanthochromic after centrifugation.

Because of these findings, a cerebral angiography study was scheduled; it demonstrated an arterial blush corresponding to the meningioma. No aneurysm or arteriovenous malformation was observed (Fig. 1 right). Based on these findings, our diagnosis was olfactory groove meningioma associated with an idiopathic SAH. Other investigations included MR imaging with and without addition of a contrast agent. It was surprising that the tumor presented with the features of a cavernoma: hyperintense and heterogeneous central signal on T₁- and T₂-weighted MR images and hypointense peripheral rim (Fig. 2). Nevertheless, the lesion was extraaxial and arose from the olfactory groove. After these MR imaging findings, the initial diagnosis became doubtful, but no other hypothesis was advanced.

Operation and Postoperative Course. Surgical removal of the lesion via a right subfrontopterional approach was performed by the senior author (T.C.). The tumor was actually extraaxial and arose from the dura mater of the olfactory groove. Moreover, the consistency of the lesion was that of a typical meningioma. Nevertheless, the color of the resected tumor was heterogeneous, with yellow, green, and brown areas, which are typically seen in a cavernoma after repeated bleeding episodes. This led us to suspect the presence of an associated cavernoma, as suggested by the MR imaging features. A mulberry lesion within the meningioma and seated on the dura mater was seen during surgery and was radically removed along with the tumor. As seen in Fig. 3, a postoperative MR image, after the dissection the left olfactory tract was intact. The patient had no postoperative complications and was discharged from the hospital on the 7th day after surgery.

Histopathological Findings. Microscopic sections confirmed the association of a transitional type meningioma with a cavernoma. There were lobules of meningothelial,

Abbreviations used in this paper: CT = computerized tomography; MR = magnetic resonance; SAH = subarachnoid hemorrhage. 

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elongated cells, but neither atypical nor malignant features were identified. Numerous hemosiderin deposits into the stroma were seen. At the periphery of the meningioma there were numerous channels with endothelial cells. These channels had moderately thick walls, large lumina, and were surrounded only by connective tissue (Fig. 4).

**Discussion**

*Intracranial Hemorrhage, Meningioma, and Cavernoma*

It is well known that brain tumors of different types may be revealed by hemorrhage. The tumoral origin of intracerebral hematomas is not a rare event, occurring in approximately 4.4% of cases in a large series over a 30-year period. In the same paper, the authors found that onset of hematoma is more common in anaplastic glioma, followed by meningioma, than in other types of brain neoplasms. In meningiomas presenting with intracerebral bleeding, the site of this bleeding may be located within the meningioma itself, in the subdural or subarachnoid spaces. The lesion may be stretching communicating veins in the subdural space and making them more susceptible to rupture or hemorrhage within the meningioma. This phenomenon may extend out of the tumor into the subdural space, or even into the brain parenchyma. Extensive intratumoral infarction may also be an important process leading to peritumoral hemorrhage in meningiomas. In our case, data from the CT scan and particularly from cerebral angiography, which only displayed the meningiomatous blush, led us logically to suspect an olfactory groove meningioma.

On the other hand, cavernomas are also known to be revealed by intracerebral bleeding and sometimes by an SAH, with a general annual hemorrhage rate of 1.3%; 0.6% in patients with no previous bleeding episode and 4.5% in patients who experienced one bleeding episode. Clinical presentation with SAH is rarely mentioned, and generally occurs when the cavernoma is located in an intraxial–subarachnoid location or in the ventricle.

Once the MR imaging study was performed, the diagnosis of meningioma did not seem to be completely adequate. Of course, an angiographically confirmed meningiomatous blush was present, but the MR imaging studies demonstrating the classic honeycomb core surrounded by a rim with low signal intensity on T2-weighted images supported the diagnosis of a possible cavernoma associated with the meningioma. One hypothesis explaining the rare but not exceptional occurrence of hemorrhage associated with meningioma could be the presence of an undiagnosed cavernoma associated with the tumor. Most of the published cases of meningiomas revealed by hemorrhage were reported before the MR imaging modality was widely available. The cavernomas therefore could not be suspected and perhaps were undiagnosed during surgery or pathological examination. Nevertheless, this hypothesis is very uncertain and needs to be confirmed with other cases similar to ours.

**Association Between Meningiomas and Cavernomas**

Very few reports mention the coexistence of a meningioma and cavernoma in the same patient. Furthermore, some dural cavernomas may mimic a meningioma. To our knowledge, our case represents the first report of a cavernoma located within a meningioma.

![Fig. 1. Left: Admission axial CT scan demonstrating a highly probable olfactory groove meningioma. A slight edema is visible surrounding the lesion. Right: Cerebral angiography study demonstrating the vascular blush around the tumor. This aspect was consistent with the hypothesis that the lesion could be an olfactory groove meningioma. The diagnosis became highly probable.](image1)

![Fig. 2. Preoperative axial T2-weighted MR image. In this view, the diagnosis of cavernoma is possible because of the lesion’s typical appearance on neuroimaging: the hyperintense and heterogeneous central signal and the peripheral rim with hypointense signal.](image2)

![Fig. 3. Postoperative sagittal MR image demonstrating total removal of the lesion (T1-weighted image with Gd contrast injection).](image3)
ly reported a meningioma and associated cavernous hemangio-

Fig. 4. Photomicrograph showing the association of a meningioma (1) and a cavernoma (2) in the same resected lesion. H & E, original magnification × 400.

malignant as a parasellar tumor. The cavernoma was nevertheless not located within the meningioma. Considering the incidences of these two tumors (400/100,000 for cavernomas in the very large series of Del Curling and associates and ~2.3/100,000 for meningiomas in Rohringer, et al.), the risk of presenting with this association may be estimated at approximately one per 10 million individuals.

Roux-Vaillard and colleagues report three cases of meningioma revealed by hemorrhage, and one of them (Case 3) is particularly interesting: a left frontotemporal meningioma was completely removed and results of pathological examination showed a “meningioma without atypical features or mitoses, but with the presence of a vascular re-

arrangement looking like a cavernous angioma.” This is the only paper in our literature review in which such an association may be suspected. Nevertheless, there was no clear evidence of cavernoma in that report. In our case, pathological examination provided evidence that the lesion was neither intrinsic to vascular channels of the meningioma nor an angiomatous meningioma.

Extraaxial Cavernomas

Another singularity of this case report is the site of the cavernoma. Because of its location with the meningioma on the olfactory groove, this cavernoma must be considered an extraaxial malformation. Most lesions of this sort arise from the cavernous sinus and spread to the temporal fossa, the sellar or parasellar region, or even to the orbit. Reports of tumors in other sites, including the cerebellar ten-

torium, the torcular herophili, and the petrosal sinus, were found in the literature. Meyer, et al., emphasized that extra-

This condition has been reported. To our knowledge, the ol-

factory groove has never been described as a possible location for an extraaxial cavernoma.

Conclusions

We present a unique case of a typical olfactory groove meningioma associated with a cavernoma. This is a very unusual case for several reasons: first because of the rarity of extraaxial cavernomas (which mostly arise from the cavernous sinus), second because of the extraordinary cavernoma–meningioma association, and third because of the patient’s presentation with an SAH. Although it is rare, this association is thus possible, and radical excision should be performed to avoid other bleeding episodes and for definitive cure of the patient.

References

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