Intracranial arteriovenous malformation and dural arteriovenous fistula embedded in a meningioma—case report and review of the literature☆☆☆

Moujahed Labidi *, Geneviève Lapointe
Neurosurgery Division, Neurological Sciences Department, CHU de Québec, Hôpital de l’Enfant-Jésus

A R T I C L E   I N F O

Article history:
Received 16 May 2015
Revised 8 August 2015
Accepted 17 August 2015

Keywords:
Meningioma
Arteriovenous malformation

A B S T R A C T

The association between a vascular malformation and a meningioma is a rare occurrence intracranially. We document the case of a 59-year-old man who presented with a right parietal extra-axial mass with headaches and seizures. Hemangiopericytoma was initially suspected on the basis of an unusual vascular pattern and rapid lesion progression. Angiography revealed abnormal vessels and an early draining vein associated with the mass. Arterial feeders were primarily from the middle cerebral artery with discrete contribution from both middle carotid arteries (ECA). Incidentally, a right posterior communicating artery aneurysms measuring 1.7 mm was also noted.

1. Introduction

Meningiomas are among the most frequently diagnosed primary intracranial tumors, accounting for 24–30% of primary central nervous system tumors. However, the association between meningeal tumors and a vascular malformation has been reported in only a few cases (Supplemental Table 1) [4], with the majority of vascular malformations being dural arteriovenous fistulas (dAVF) [5,6]. We report a case of concurrent pial arteriovenous malformation (AVM) and dAVF embedded in a meningioma and discuss common pathophysiological pathways between these lesions.

2. Case report

A 59-year-old man was referred to our department for a right parietal lesion. Previous medical history was significant for atrial fibrillation treated with warfarin and a history of head trauma sustained in a motor vehicle accident as a passenger, 5 years prior. At that time, an acute right subdural hematoma (SDH) and right convexity and Sylvian fissure subarachnoid hemorrhage were treated conservatively. At the one-month follow-up, the patient was asymptomatic and the computed tomographic (CT) scan had normalized.

In the months prior to his second referral, the patient had two episodes of left partial tonic-clonic seizures. These led to the identification of a chronic right-sided SDH measuring less than a centimeter. This SDH was treated conservatively with phenytoin and reversal of anticoagulation. A second head CT, obtained one month after the initial scan, documented resolution of the SDH but also revealed a right parietal extra-axial mass. Clinically, the patient began to present intermittent headaches and complained of paresthesias affecting his left hand without any recurrence of seizure. No focal deficit was found on neurological examination except hypoesthesia of the left hand. Magnetic resonance imaging (MRI) showed a rounded parietal mass, hypointense on T1 and hyperintense on T2 and on fluid attenuated inversion recovery (FLAIR) images (Fig. 1A–C). Enhancement following gadolinium injection was inhomogeneous and susceptibility-weighted images (SWI) confirmed prior hemorrhages with signal changes compatible with local hemosiderosis. In comparison to the CT scan previously obtained, the lesion appeared to be rapidly progressing. Additionally, in the adjacent gyri, multiple tortuous small vessels were identified. Suspecting a hemangiopericytoma, we obtained an angiography (Fig. 1D–F), which demonstrated an early draining vein and an area of abnormal blush. Arterial feeders came predominantly from the right internal carotid artery (ICA) with smaller contributions from the left ICA and bilateral external carotid arteries (ECA). Incidentally, a right posterior communicating artery aneurysms measuring 1.7 mm was also noted.

☆☆☆

Funding: Local funds.

© 2015 The Authors. Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).
In the light of a growing lesion suggestive of hemangiopericytoma, surgical resection was undertaken. A right parietal craniotomy revealed a dural-based lesion compatible with a meningioma, which was resected en-bloc (Fig. 2A). A dAVF draining in an arterialized vein of Trolard was also identified and disconnected. Following the disconnection of the dAVF however, the vein of Trolard remained arterialized. This led to the identification of a vascular lesion underneath the meningioma based on the pia mater (Fig. 2B). This lesion proved to be an AVM, with a distinct nidus that was dissected from the underlying parenchyma.

Postoperative outcome was favorable, but hand hypoesthesias persisted. There were additional ictal episodes, but these subsided with the addition of levetiracetam. A postoperative MRI confirmed the total removal of the tumor and angiography demonstrated exclusion of the AVM and dAVF. Histology and immunohistochemistry confirmed the diagnosis of meningioma, although a post-traumatic meningothelial hyperplasia was entertained in the differential (Fig. 3A and B). A negative CD34 immunohistochemistry ruled out hemangiopericytoma (Fig. 3C). The lesion proliferative index was low and vascular endothelial growth factor (VEGF) was not expressed in the tumor. The histology of the vascular lesion was consistent with an AVM (Fig. 4). At 3 months, MRI and CT-angiography were repeated and showed no residual tumor or vascular malformation. Clinically, there was no recurrence of seizures and paresthesias subsided.
3. Discussion

We report a case of concurrent and anatomically associated meningioma, dAVF and pial AVM. These presented 5 years following subdural and subarachnoid hemorrhages secondary to head trauma. 11 previous cases of AVMs associated with meningiomas (9 cases) or hemangiopericytomas (2 cases) have been previously reported (Supplemental Table 1). Including the present case, the tumoral and vascular lesions were contiguous in only 6 cases and only 2 patients presented intracranial bleeding before surgical intervention. AVMs and meningiomas were majoritarily located on the convexity. Including the present case, 3 patients had a dAVF in association with an AVM.

This rare association between an AVM and a meningioma and their shared vascularity raises questions as to their respective pathophysiology; has one led to the development of the other or do these lesions share common pathogenic pathways. In fact, and although AVMs are thought to be congenital lesions, there is experimental and clinical evidence that abnormal angiogenesis and vascular remodeling occur in adult patients. Angiogenesis is controlled by a number of factors, including VEGF. There is increased expression of VEGF in the vascular walls of AVMs and it has been associated with increased angiogenesis and peritumoral edema in meningiomas [1]. Although VEGF was not expressed in the meningioma described in the present case, previous authors have postulated that increased vascularity and cerebral blood flow associated with meningiomas and hemangiopericytomas may also be a factor in the development and growth of vascular malformations. The concurrent finding of a dAVF must also be weighted in the pathophysiology. It has been previously reported that meningiomas and other CNS tumors may be associated with as many as 4.6% of dAVF [7]. In many of those cases however (40.9%), tumors were associated with significant venous outflow obstruction. In comparison, there was no venous obstruction in 10 out of 11 cases of AVM associated with a dural lesion, suggesting a lesser role of venous hypertension in the concurrent development of AVM and meningioma.

Previous trauma and consequent hemorrhages in the subarachnoid and subdural compartments may well have contributed to the induction of an inflammatory environment. It has been previously postulated that such chronic inflammation in the post-traumatic setting may lead to the formation of meningiomas. In AVMs, inflammatory mechanisms have been involved in both lesion progression and rupture [3]. In addition to the inflammatory changes induced by trauma, one may speculate that the initial hemorrhages may have been consequent to damage to cortical vessels in the parietal lobe. Fistulous connection between cortical and dural vessels may have developed and persisted during the inflammatory and healing phases. Nevertheless, no vascular imaging was done following the initial trauma and thus, the possibility that the AVM was already present before the trauma cannot be ruled out.

Clinically, the presence of a vascular malformation may be difficult to diagnose, owing to the hypervascularity of the meningeal tumor [2]. A preoperative angiogram may help in the identification of arterial feeders and draining veins. The treatment of either lesion should take into account the attributable symptomatology and natural history of each lesion [4]. The
differential diagnosis includes meningioangiomatosis, a rare and benign hamartomatous lesion that may be associated with both meningioma and vascular malformation. Typically, these are extra-axial plaque-like lesions that present with seizures. Histopathology, in the absence of the typical cortical hypervascularity and perivascular spindle cells, allowed us to exclude this diagnosis. Likewise, the diagnosis of meningioma was preferred to that of post-traumatic meningothelial hyperplasia because of the cellularity of the lesion, the dural invasion and macroscopic appearance.

4. Conclusion

The association between vascular malformations and meningiomas is a rare occurrence intracranially. Such cases illustrate the pathogenic roles of angiogenesis and inflammation that is common to these lesions.

Conflicts of Interest

The authors report no conflict of interest.

Supplementary data to this article can be found online at http://dx.doi.org/10.1016/j.inat.2015.08.001.

Acknowledgments

The authors would like to thank Ms. Élène Porter for the graphical work related to this paper and neuropathologists Dr Stephan Saikali and Dr Peter V. Gould.

References