LETTER / neurology

Coexistence of intracranial meningiomas and vascular malformations: A fortuitous association or direct relationship?


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Meningiomas are the most common primary intracranial tumours after the gliomas [1]. While they are usually associated with other intracranial tumours, falling generally into the neurofibromatosis category, their association with vascular malformations is still little reported and is probably underestimated.

Through our two observations, one associating a meningioma of the foramen magnum with a frontotemporal cerebral arteriovenous malformation (AVM), and the other, a meningioma of the posterior surface of the temporal bone with a cerebellar cavernoma, we shall discuss the various hypotheses that might explain this association.

1. Observations

1.1. Case n° 1

This female patient, aged 59 had had an epileptic seizure 30 years ago followed by left side heaviness. Cerebral angiography performed at the time revealed a large right frontotemporal cerebral AVM, supplied by branches of the middle cerebral artery, combined with multiple varicosities. The rest of the examination did not detect any abnormality. Since she refused all treatment options, the patient was clinically monitored; no neurological sign came to light for 23 years. In 2000, the patient presented balance disorders followed by the onset of left side heaviness. Magnetic resonance imaging
(MRI) of the brain objectified the middle cerebral AVM, which showed no sign of bleeding, with an extra-axial process from the foramen magnum which was roughly oval in shape, situated to the right, exhibiting isosignal intensity in T1 and T2 -weighted sequences and showing intense and uniform contrast pushing into the inferior end of the pons and medulla (Fig. 1). After total surgical exeresis of the process, anatomopathological examination confirmed its richly vascularised meningotheial nature. A follow-up MRI performed in 2005, revealed recurrence of the meningoia as a trilobed mass, isointense in T1 and T2 -weighted images with high uptake of the contrast agent. Since the patient refused further surgical treatment, she underwent stereotactic radiotherapy (Fig. 1). A dose of 15 Gy was prescribed to the periphery of the meningoia and embolisation of the cerebral AVM was suggested.

1.2. Case n° 2
This 71-year-old patient had had balance disorders associated with vertigo for 3 months. Cerebral MRI objectified two processes (Fig. 2). The first was an extra-axial process of the right cerebellopontine angle, widely implanted in the dura mater and connected at an obtuse angle to the posterior surface of the temporal bone. This process was of isosignal intensity in T1 -weighted images, hyperintense with T2 -weighting, with intense uptake of the contrast agent following injection of the contrast agent, compatible with a meningoia.

The second process was at the bottom of a cerebellar fissure of the left hemisphere, was roughly rounded, and comprised a heterogeneous central zone with areas of hyposignal and hypersignal surrounded by a hyposignal ring, compatible with a cavernoma. Since the patient opted for stereotactic radiotherapy, the meningoia was given a peripheral dose of 15 Gy (Fig. 2) and open surgery of the cavernoma offered to the patient.

2. Discussion
The simultaneous presence in a single individual of a primary tumour of the brain and a cerebral AVM is rare. In 1960, Fine et al. [2] were the first to report the coexistence of a right parietal cerebral AVM and an intraventricular oligodendroglia. About 50 cases of cerebral AVM combined with cerebral tumours have been reported in the literature [3], 14 of which were intracranial meningoias, including our observations.

Classification has been suggested depending on the location of the two lesions [4].
Type I are lesions separated from each other and situated in different anatomical locations, type II are cerebral AVMs contiguous with the tumoral mass with or without a clear cleavage plane from the latter, while type III are cerebral AVMs adjacent to the tumour within the same gyrus or lobe. Our first case is type I in view of the fact that the cerebral AVM is situated in the right cerebral hemisphere and the meningoia affects the foramen magnum. Several hypotheses have been aired to explain the coexistence of these two neoplasms within the same parenchyma [4,5,6]:

- cerebral AVM can induce tumour formation;
- the tumour can cause development of an AVM, or at least increase blood flow from a cerebral AVM, hitherto quiescent;
- the two lesions could be induced by a third environmental factor.

Cushing and Eisenhardt [1] suggested that leptomeningeal tumours could result from chronic irritation of the arachnoid cells by a pathological process of some type, including cerebral AVM, by increasing the blood flow in the arteries supplying it. Other authors have incriminated humoral factors such as tumour angiogenesis factor secreted by the cerebral AVM and triggering appearance of the tumour [7].

An added genetic factor may be implicated: Kasantikul and Brown reported two observations of meningoimatoses without patent signs of neurofibromatosis, including one associating a meningoia and cerebral AVM. They nevertheless raised the possibility of a tumour and cerebral AVM being associated in the context of neurofibromatosis, for which the clinical, paraclinical and genetic criteria need to be found [8]. We did not observe any phacomatos in our two patients.

It has been reported that cerebral AVM may not be recognised on imaging within the meningoia owing to its thrombosis, the intracranial hypertension or the local pressure of a haematoma [7]. A good study multiplying the radiological investigations (CT angiography, MRI with angiography sequences, 3D angiography) could highlight these vascular lesions and offer approaches to treatment.

The association of a meningoia with an aneurysm is the most frequently reported vascular malformation involving meningoias: 42 cases (excluding iatrogenic aneurysms) have been described, including one case of an intratumoral association. Two cases were discovered when postoperative subarachnoid haemorrhage occurred far from the area of the exeresis, implicating decompression resulting from eradication of the meningoia [9]. However, since the appearance of most aneurysms is at the expense of the arteries supplying the meningoia, the increase in cerebral blood flow in these vessels has been considered a contributory factor, especially if there is arterial hypertension [10]. Most demands are on the middle cerebral artery followed by the internal carotid artery. Ten or so cases have been reported of non-traumatic aneurysm of the middle meningeal artery associated with meningoias, most often of the temporal convexity and base [11].

The association of a meningoia with a dural arteriovenous fistula (DAVF) is also rare. These meningoias are usually inserted near the dural sinuses or arise from within their walls, resulting in obstruction or invasion of the sinus lumen, thus causing the development of the DAVF [12]. Obstruction of the sinus is not a prerequisite according to Ahn for the formation of a DAVF [13].

Rare cases have been described of cavernomas associated with meningoias [14,15]. These observations implicate genetic factors and abnormalities in development. The introduction of a polymavirus into the brain of a rodent caused the formation of cavernomas within its parenchyma [16]. For Kunishio [16], their coexistence does not seem to be fortuitous given the common mesodermal origin of the
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Figure 1. T₁-weighted cerebral MRI in the three spatial planes after injection of the contrast agent: A, B: Large right frontotemporal cerebral arteriovenous malformation (AVM). C: Extra-axial trilobed process from the foramen magnum with high uptake of the contrast agent. D: Large right frontotemporal cerebral AVM. E: Concomitant view of the temporal portion of the cerebral AVM and the retroclival portion of the meningioma. F: Dosimetric plan for stereotactic radiotherapy of the meningioma.
Figure 2.  T1-weighted cerebral MRI axial and coronal slices after injection of the contrast agent: A and B: Hyperintense extra-axial process of the right cerebellopontine angle largely implanted in the dura mater and lying against the posterior surface of the temporal bone and the tentorium cerebelli. C and D: Left cerebellar process at the bottom of a cerebellar fissure comprising a heterogeneous central zone surrounded by a hyposignal ring. E: Dosimetric plan for the stereotactic radiotherapy of the meningioma.
two neoplasms. Holland [14] reported that a cavernoma was discovered following bleeding on the day following surgery of a meningioma of the sphenoid jugum, which was investigated with slices passing solely through the sellar region, whereas the unknown cavernoma was located in the posterior part of the right internal capsule.

3. Conclusion

Meningioma associated with vascular malformation is a rarely reported entity. It should be considered preoperatively so as to complete the neuroradiological investigations necessary for appropriate therapeutic management.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

References