CASE REPORT

Hemorrhagic collision metastasis in a cerebral arteriovenous malformation

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SUMMARY

A 26-year-old patient with recurrent choriocarcinoma of the testis presented with headache and progressive left homonymous hemianopsia. On initial MRI a grade 4 arteriovenous malformation (AVM) was identified in the right occipital lobe, which was further characterized by catheter angiography. Continued worsening of the headache in the following days prompted a follow-up MRI, which revealed a new T2 hypointense nodule and adjacent vasogenic edema in the periphery of the AVM. A follow-up MRI showed a marked increase in the size of the nodule with intrinsic enhancement and worsening perilesional edema. Based on the imaging evolution, the nodule was diagnosed as a metastasis and the patient was started on chemotherapy and radiotherapy. One week after the MRI he developed a sudden hemorrhage within the mass requiring decompression craniectomy and resection of both AVM and tumor. The histopathology of the resected mass confirmed the diagnosis of choriocarcinoma metastasis to the AVM.

BACKGROUND

Collision metastasis within pre-existing brain arteriovenous malformations (AVMs) is a rare occurrence. To our knowledge, this is the first such reported case of collision metastasis and AVM.

CASE PRESENTATION

A 26-year-old patient with metastatic testicular choriocarcinoma to sternal lymph nodes presented with headache and progressive right homonymous hemianopsia. Contrast-enhanced MRI revealed a region of serpiginous flow voids within the posteromedial right occipital lobe, consistent with an arteriovenous malformation (AVM). A catheter angiogram showed an AVM with a 5 cm×3 cm nidus, supplied by the right middle cerebral and posterior cerebral arteries and draining into the cortical veins (Spetzler–Martin grade 4 AVM). Follow-up MRI within a month showed an adjacent 1 cm rim-enhancing mass with perilesional edema, suggesting metastasis rather than AVM hemorrhage (figure 1A, B).

The patient continued to have worsening headache in the following days and 3 weeks later a positron emission tomography (PET) scan and follow-up MRI was obtained. The PET scan showed no uptake in the brain lesion or in the known metastases. On the MRI, the nodule increased in size to 4.8 cm×3.0 cm with

perilesional edema and showed contrast enhancement (figure 1C, D). After progression of disease was established, the patient initially opted for chemoradiation treatment over surgical or endovascular treatment. Intensive radiation treatment and chemotherapy was instituted for the metastatic choriocarcinoma. Unfortunately, 5 days after initiating treatment the tumor hemorrhaged spontaneously with subsequent mass effect and herniation syndrome.

TREATMENT

The patient underwent emergency right-sided decompressive craniectomy and resection of the metastasis and AVM. Due to the intimate association of the tumor with the posterolateral border of the AVM, both required gross total resection. Histopathology confirmed hemorrhagic choriocarcinoma and AVM.

INVESTIGATIONS

The resected specimen contained a well-vascularized hemorrhagic and necrotic mass within a predominantly gliotic brain parenchyma containing the pial AVM and arterialized veins. The neoplastic cells were bizarre-appearing large cells with a large irregular nucleus containing very large nucleoli, coarse chromatin and had abundant basophilic to vacuolated cytoplasm. Scattered multinucleated syncytiotrophoblastic forms were also present. There were no intravascular tumor cells. The histopathology features matched with his orchiectomy specimen, confirming the source of the primary tumor (figure 2).

DIFFERENTIAL DIAGNOSIS

Ruptured AVM versus metastatic choriocarcinoma as a collision metastasis to the AVM.

OUTCOME AND FOLLOW-UP

The patient tolerated surgical treatment with persistent visual field deficits at the time of discharge to a skilled nursing facility. Because of this metastasis, his overall prognosis remains guarded and he is being considered for further chemotherapy by his oncology team.

DISCUSSION

Hematogenous metastatic cells require a normal capillary system and venules for successful anchorage to the endothelium and subsequent

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Figure 1 (A) Axial T2W image on initial MRI showing flow voids in the arteriovenous malformation (AVM) within the right occipital lobe (arrow). (B) Axial T2W image on the first follow-up MRI showing a T2 hypointense nodule in the lateral aspect of the AVM (arrowhead), with increase in the edema in the occipital white matter. (C) Axial T2W image from second follow-up MRI showing progressive enlargement of the nodule, now appearing more heterogeneous in signal intensity. (D) Axial T1W post-contrast image demonstrating enhancement within the nodule which is highly suggestive of a neoplastic process in this case.

proliferation. The blood vessels in the AVMs have a thickened medial layer of the arterial wall due to hyalinization, lacking normal intervening capillary network and arterialization of the draining veins. Due to high-flow shunting, there is gliosis of the intervening and adjacent brain parenchyma. These adverse conditions drastically reduce the chances of successful implantation of the metastatic cells within or in the vicinity of the AVM.¹ Metastatic choriocarcinoma can cause intracranial hemorrhage which may be the initial symptom of gestational trophoblastic disease due to its propensity for early hematogenous metastasis. Occasionally, such lesions may be mistaken for a ruptured AVM.² Testicular choriocarcinoma is a rare non-seminomatous germ cell tumor, accounting for 1.5% of all testicular neoplasms.^{3,4} Metastasis to the brain occurs by additional arterial

tumor emboli arising from the pulmonary metastases. Histologically, the tumor metastasis consists of cytotrophoblasts and syncytiotrophoblasts which often partially occlude the vascular lumen. The syncytiotrophoblasts have an innate propensity to invade the endothelium and extend into the perivascular spaces, thereby weakening the vessel wall and causing fusiform and saccular 'oncotic' aneurysms in the distal intracranial branches.⁵⁻⁷ In the case presented here, no intranidal or adjacent aneurysms were identified on the angiogram; however, given the rapid growth pattern exhibited by the metastasis following the angiogram, this possibility cannot be excluded. The earlier MRI exhibited a low T2 signal nodule which presented a diagnostic quandary as to whether this represented a spontaneous hemorrhage or thrombosis from the AVM or a

Figure 2 Metastatic choriocarcinoma adjacent to abnormal vessel (open arrow). Islands of viable carcinoma with abundant necrotic neoplasm are noted within extensive hemorrhage. There were no intravascular tumor cells. Insert: High power view of carcinoma cells showing multinucleated syncytiotrophoblast component (arrow) and smaller uninuclear cytotrophoblast cells (dotted arrow). β -Human chorionic gonadotropin immunohistochemical stain positive (not shown).

hemorrhagic metastasis. The pattern of evolution of this nodule on follow-up imaging led us to suspect metastasis. It is plausible that the tumor deposition occurred in the normal brain parenchyma in close proximity rather than within the AVM itself, and was entirely uninfluenced by the local hemodynamic changes brought about by the AVM. Brain parenchyma adjacent to AVMs is typically vasodilated to counteract any local steal phenomenon. This also may have predisposed the patient to hematogenous metastatic spread adjacent to the AVM. However, this fine distinction could not be prospectively made on MRI or examination of the pathology specimen. The hemodynamic

Learning points

- ▶ Hemorrhagic metastasis to the brain from highly vascular tumors such as choriocarcinoma is common.
- ▶ Albeit rare, collision metastasis to an arteriovenous malformation (AVM) can occur.
- ▶ If there is diagnostic uncertainty as to whether or not a metastasis exists, very close follow-up imaging should be done to evaluate for changes in the lesion morphology. In cases where collision metastasis is highly suspected, early resection with or without embolization of the AVM should be carried out.

idiosyncrasies prevailing in and around the AVM make the location of this metastasis intriguing and unprecedented.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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