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Case Report

The coexistence of oligo-astrocytoma, arteriovenous malformation and aneurysm: a rare case

Jing Zhang[1*], Yanlin Song[2*], Min He[1*], Chenjing Zhu[2*], Jianchao Wang[3], Jieke Liu[4], Zhiyong Liu[1], Qingqing Ren[1], Baoyin Shan[1], Yunhui Zeng[1], Xuelei Ma[2], Jianguo Xu[1]

[1]Department of Neurosurgery, [2]Cancer Center, Departments of [3]Pathology, [4]Radiology, West China Hospital, Sichuan University, Chengdu 610041, PR China. [*]Equal contributors.

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Abstract: This report is of a rare case of mixed oligo-astrocytoma and arteriovenous malformation (AVM) associated with an aneurysm in a 50-year-old man, who presented with a progressive history of headache and vomiting due to acute intracranial hemorrhage. Initial images were suggestive of a right frontal AVM and an aneurysm on the left anterior cerebral artery (ACA) for which a right frontotemporal craniotomy was performed. Intraoperatively, a gross total resection of the AVM was achieved and then the aneurysm was clipped. After surgery, histopathological examination demonstrated oligo-astrocytoma in the resected lesion of AVM. The clinical, radiological, and operative data were reviewed, so were the histological findings. To the authors' knowledge, this is the first reported case with glioma, AVM and aneurysm. The association of tumors and vascular malformations should always be taken into consideration. We suggest that, in selected cases, preoperative angiography should be performed in patients with brain tumor, and that preoperative MRI scan and postoperative accurate histopathological evaluation are required to exclude tumors in patients with AVM. So far, whether intermixed or closely related tumors and vascular malformations are combined or represent two entities remains unclear. Further studies of more such patients are needed to explain such an association.

Keywords: Glioma, AVM, aneurysm, case reports

Introduction

Gliomas are the most common type of primary intracranial tumor [1]. Due to the neovascularization in tumor, patients may present with intratumoral hemorrhage [2]. Intratumoral hemorrhage is reported to occur in 3.7-7.2% of gliomas, mainly in high grade gliomas [3]. Arteriovenous malformations (AVMs) compose tortuous arteries and veins, and lack an intervening capillary bed [4]. Yearly hemorrhage rates in patients with AVM range from 2% to 32.6% [5]. Sudden onset of neurological symptoms is conventionally thought to be due to vascular diseases, but intratumoral hemorrhage may be an important differential diagnosis [6]. Intracranial coexistence of glioma and AVM is very rare, and data are limited to case reports [7-11]. Acute hemorrhage in such patients may due to the AVM or glioma. We present a 50-year-old man with mixed oligo-astrocytoma and AVM in the right frontal lobe and aneurysm on the left anterior cerebral artery (ACA), presenting with intracranial hemorrhage. This is the first reported case with those three entities.

Case report

History

A 50-year-old man presented in our hospital with a three-day history of progressive headache with vomiting. No history of recent trauma was found. On admission, his blood pressure was 147/98 mmHg and the Glasgow Coma Scale (GCS) score was 15. The initial CT-imaging revealed a right-sided frontal hybrid density mass lesion with perifocal edema, swelling of the brain, slight compression of the right lateral ventricle and subarachnoid hemorrhage (**Figure 1A**). Additional computed tomographic angiography (CTA) showed no apparent engorged and enhanced vascular mass in the right frontal hematoma area, but revealed a small projec-

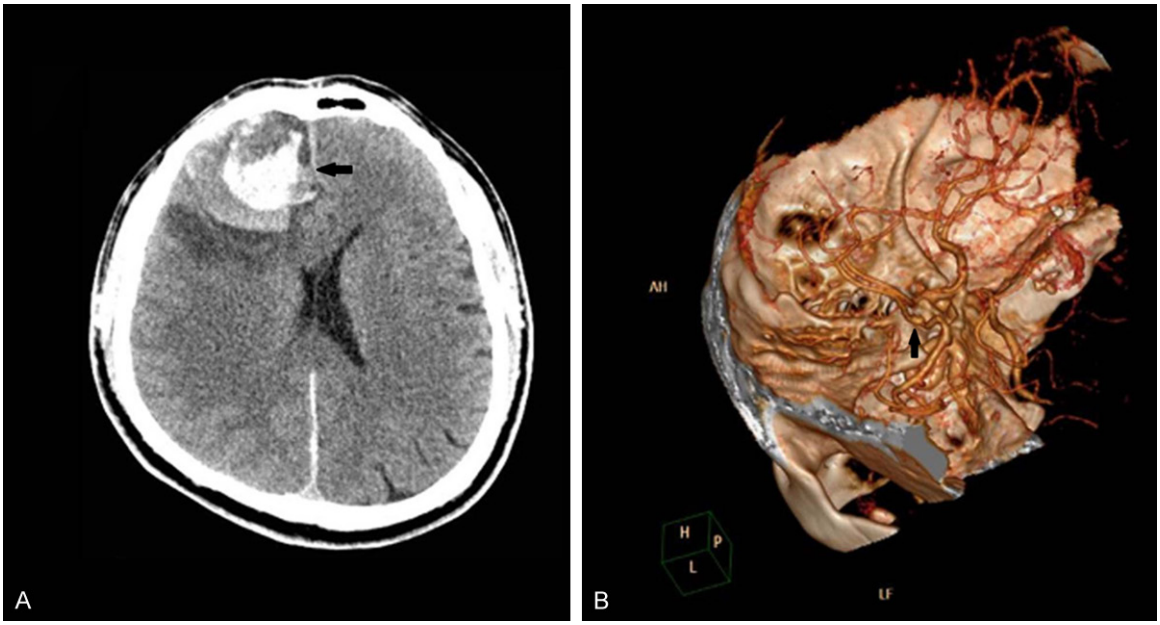

Figure 1. CT scan and computed tomographic angiography (CTA) on admission. CT scan showed a right-sided frontal hybrid density mass lesion (arrow) with perifocal edema, swelling of the brain, slight compression of the right lateral ventricle and subarachnoid hemorrhage (A). CTA revealed a small projection (arrow) on the A1 segment of the left anterior cerebral artery (ACA) (B).

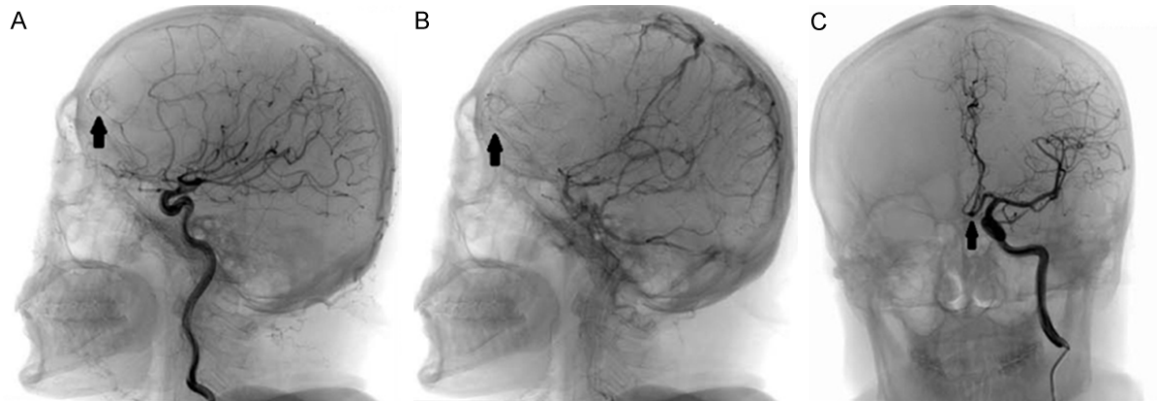

Figure 2. Diagnostic digital subtraction angiography (DSA). Lateral views of a diagnostic right internal carotid artery (ICA) DSA demonstrated a right frontal AVM (arrow) supplied by the right polar frontal artery (A and B). The postero-anterior view of a diagnostic left ICA DSA showed an aneurysm (arrow) at the end of the A1 segment of the left anterior cerebral artery (ACA) (C).

tion on the A1 segment of the left ACA (**Figure 1B**). Then, the diagnostic cerebral digital subtraction angiography (DSA) demonstrated a right frontal AVM supplied by the right polar frontal artery and confirmed the presence of aneurysm at the end of the A1 segment of the left ACA (**Figure 2**).

Operation

The decision was made to perform a surgical procedure. After administration of general anesthesia, the patient underwent right fronto-temporal craniotomy, which revealed abnormal vascular mass and dark red blood. After isolating the border of the vascular malformations, a gross total resection of the lesion was achieved and old hematomas were cleared. The resected lesion was sent for histological examination. Afterwards, optic nerves, optic chiasma, internal carotid artery and ACA were isolated, and the aneurysm at the end of the A1 segment of the left ACA was exposed. The

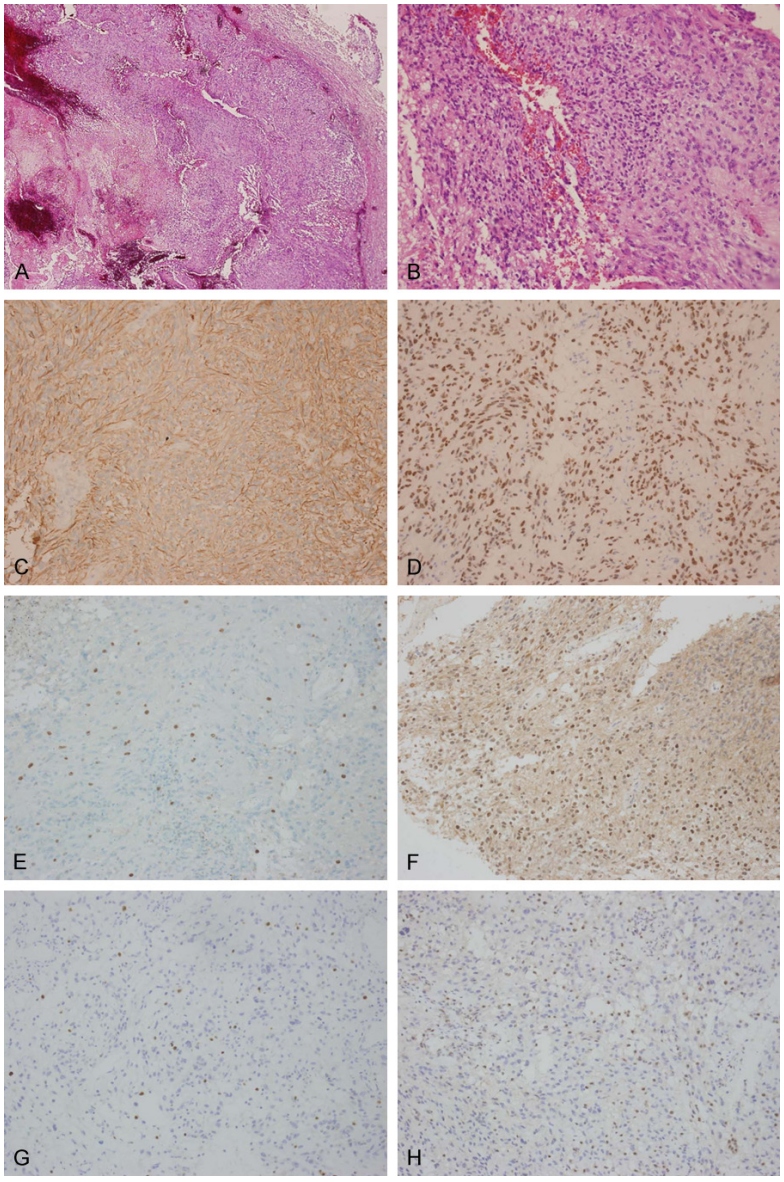

Figure 3. Histopathology of oligo-astrocytoma. Neoplastic infiltration (H&E stain, original magnification ×40) (A). Neoplastic infiltration (H&E stain, original magnification ×200) (B). Immunohistochemical staining revealed that tumor cells expressed GFAP (C), Oligo2 (D), Ki-67 (E), P53 (F), Topo II (G) and MGMT (H) (C-H original magnification ×200).

aneurysm extended anterosuperiorly, and its wall was thin. Then the aneurysm was clipped. The patient remained intubated and ventilated for 24 h. The patient's clinical condition was stable and he was transferred to our rehabilitation ward 8 days after the surgery.

Histological examination

The histological examination showed diffuse and infiltrative oligodendrocyte and astrocyte tumor, with necrosis, microvascular proliferation and few nuclear mitosis. The immunohistochemistry showed: GFAP (+), Oligo2 (+), IDH1 (-), Ki-67 (+, 15%), P53 (+), PTEN (-), Topo II (+, <5%), MMP9 (-), MGMT (+, 10%), EGFR (-) (**Figure 3**). High grade glioma (anaplastic mixed oligo-astrocytoma, WHO grade III) was proposed as a diagnosis.

Postoperative course

Four weeks after the surgery, he was admitted to the department of oncology in our hospital. MRI scan showed the margin of the surgical residual cavity was heterogeneously enhanced. He received concurrent chemoradiotherapy (intensity modulated radiation therapy plus temozolomide). Latest MRI scan (1.5 years after surgery) showed no tumor recurrence.

Discussion

We report the case of a 50-year-old man with the histological examination of an oligo-astrocytoma associated with AVM and an aneurysm. Preoperatively, he was diagnosed with AVM in the right frontal lobe and aneurysm on the left ACA. After surgery, histological examination demonstrated oligo-astrocytoma in the resected lesion of AVM. So the hemorrhage in this patient may came from the AVM or the tumor. Coexistence of AVM and glioma remains a rare event and was reported by few researchers [7-11]. In this patient, he also had an aneurysm. As far as we know, this is the first reported case with glioma, AVM and aneurysm.

Since the glioma and AVM were intermixed in this patient, and the aneurysm was on the

other side, we mainly discuss the coexistence of glioma and AVM here. When two pathological entities exist in close vicinity, a link between them may be suggested. McKinney et al. [8] provided evidence about the de novo formation of an AVM within an anaplastic oligodendroglioma in a patient with initially negative brain MR and intracranial MR angiography. The hypothesis that hyperangiogenic stimulus provided by the tumor might have initiated the development of the AVM was agreed by some researchers [8, 10, 12]. On the contrary, some researchers tend to speculate that there might be secondary neoplastic changes in the perivascular glial tissue in response to the vascular malformation [13-15]. However, cases in literature are too few to come to a conclusion regarding the etiology. On the other hand, some researchers thought that might be a fortuitous separate association [16, 17]. Whether this association is a coexistence of individual lesions or not, is still open to discussion.

Nowadays, MRI scan, especially contrast-enhanced MRI scan, can successfully diagnose most tumors, so an additional cerebral angiography to exclude vascular malformations is usually not performed [9]. In some reported cases with both brain tumor and vascular malformation, the brain tumor lesions were often found first by imaging, and the vascular malformations were found during the operation or after the pathological examination [7, 9, 11]. Thus, Ziyal et al. suggested that routine preoperative angiography, which can give us the precise information not only about the tumor itself, but also the vascularity and displacement of normal vessels, should be performed in selected cases [9]. In some cases, with the knowledge of the tumor vascularity, preoperative transarterial embolization may be performed to reduce the blood supply of the tumor, thus reducing the blood loss during surgery, which is more important in children. Besides, preoperative angiography could help with surgical plan intraoperatively. And preoperative angiography was performed routinely for brain tumors in Japan since more than 10 years ago [9].

Gmeiner et al. [7] reported an interesting case, which was glioblastoma with the appearance of AVM. The initial MRI imaging was suggestive of a high-grade glioma, but intraoperatively, the lesion resembled a vascular malformation. Initial histopathological analysis revealed an AVM. Two months later, multiple lesions were visible on MRI imaging, thus, supporting the diagnosis of malignant glioma. After reinvestigating the histopathological sections, in only 5% of the section a glioblastoma which was surrounded by an AVM-like lesion was discerned. In our reported case, the patient was also diagnosed with AVM preoperatively. But after surgery, histological examination demonstrated oligo-astrocytoma in the resected lesion of AVM. So we agree with Gmeiner et al. that postoperative accurate histopathological evaluation is required to avoid misdiagnosis and to initiate appropriate treatment strategies in patients with AVM.

Looking back on this patient, if we performed MRI scan before surgery, we might find the tumor lesion mixed with the AVM. Then the surgical removed area may be expanded to minimize the residual tumor, which was good for this patient. So we also recommend that preoperative MRI scan in AVM patients could help to exclude tumors.

In conclusion, this case is the first reported case of oligo-astrocytoma coexisting with arteriovenous malformation and an aneurysm. The association of tumors and vascular malformations should always be taken into consideration. We suggest that, in selected cases, preoperative angiography should be performed in patients with brain tumor, and that preoperative MRI scan and postoperative accurate histopathological evaluation are required to exclude tumors in patients with AVM. So far, whether intermixed or closely related tumors and vascular malformations are combined or represent two entities remains unclear. Further studies of more such patients are needed to explain such an association.

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Disclosure of conflict of interest

None.

Address correspondence to: Jianguo Xu, Department of Neurosurgery, West China Hospital, Sichuan University, No. 37, Guoxue Alley, Chengdu 610041,

PR China. Tel: +86-28-85475576; E-mail: jianguo_1229@126.com; Xuelei Ma, Cancer Center, West China Hospital, Sichuan University, No. 37, Guoxue Alley, Chengdu 610041, PR China. E-mail: drmaxuelei@gmail.com

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