

Surgical strategy in case with co-existence of malignant oligodendroglioma and arteriovenous malformation: A case report

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ABSTRACT

A brain tumor associated with an arteriovenous malformation (AVM) is very rare. A 42-year-old female presented with two separate lesions in her right frontal lobe on MRI. An angiogram diagnosed one of the lesions as an AVM. The second lesion appeared to be a tumor. Tumor removal was difficult due to bleeding from the nearby AVM, necessitating removal of the AVM and allowing complete excision of the tumor. Histopathological analysis revealed the tumor was an anaplastic oligodendroglioma. There was no recurrence of the tumor 5 year after completion of therapy. We discuss the operative strategy in case of synchronous diseases and provide a review of the literature.

Keywords: Oligodendroglioma; Arteriovenous Malformation; Co-Existence

1. INTRODUCTION

The association of a brain tumor and an arteriovenous malformation (AVM) is rare. While there have been a number of reports, few of these have reported the details of the operative strategy, particularly the approaches used based on the preoperative information of both lesions. We describe our chosen surgical strategy in a case with two such synchronous lesions and provide a review of the literature.

2. CASE REPORT

A 42-year-old previously healthy female presented

with complaints of headache and vomiting. Her level of consciousness was normal and she had no neurologic deficits. Magnetic resonance images (MRI) on admission revealed a 7 cm in diameter mass lesion that showed low signal intensity on the T1 weighted images (WI) and high signal intensity on the T2 WI in the right frontal lobe. The lesion demonstrated heterogeneous enhancement by gadolinium-diethylenetriamine penta-acetic acid (Gd-DTPA) adjacent to the Rolandic vein (**Figure 1(A)**). The T2 WI also demonstrated a second 3 cm in diameter lesion with multiple flow void signs in the right inferior frontal gyrus (**Figure 1(B)**). An angiogram showed an arteriovenous malformation (AVM) that was predominantly fed by the middle cerebral arteries and drained through the superficial frontal ascending vein and the Rolandic vein (**Figures 1(C) and (D)**). Spetzler-Martin Grade was 1 point. The angiogram also demonstrated an avascular region in the area of the former lesion. A plain computed tomography scan showed partial calcification, and a dynamic study of the methionine positron emission tomography (PET) scan presented a reduction of accumulation in the longitudinal course in the former lesion. The results of dynamic PET study indicated oligodendroglial tumor according to our study previously reported [1]. Comprehensively, these radiological findings led to a diagnosis of malignant oligodendroglial tumor associated with AVM. The patient's headache was likely caused by the large tumor, and so we planned to resect the oligodendroglioma first, followed by the AVM. For the operation, we performed a large osteoplastic frontotemporal craniotomy beyond the mid-line including both lesions, with the goal being to remove the oligodendroglioma and AVM consecutively. During this procedure, severe bleeding occurred from the

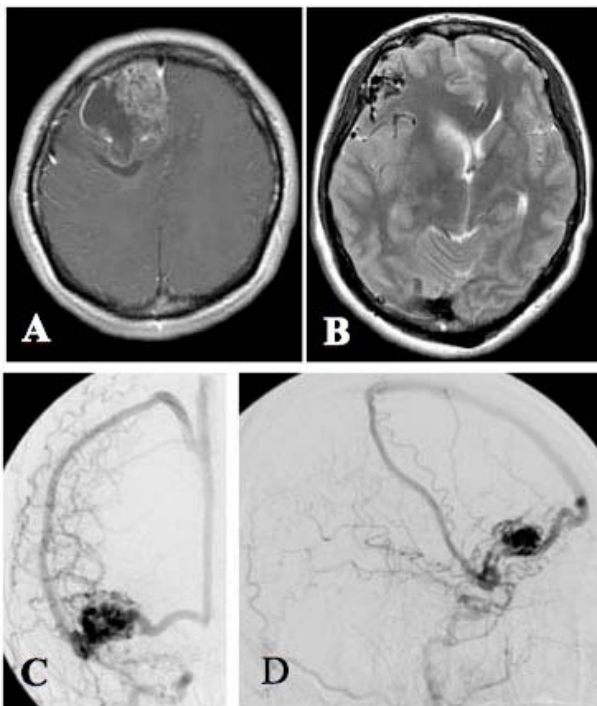


Figure 1. (A) MRI with gadolinium showing a heterogeneously enhancing mass lesion with partial cyst in the right frontal lobe. (B) The T2 weighted image showing crowded flow void signs that suggest an arterio-venous malformation in the right inferior frontal gyrus. (C), (D) The right common carotid angiogram (C: frontal view, D: lateral view) showing a nidus fed by middle cerebral artery and mainly draining to the Rolandic vein, and the anterior frontal ascending vein. The tumor area in front of Rolandic vein shows no tumor shadows.

venous sac protruding beside the superior sagittal sinus. After hemostasis was achieved and a dural incision was made, both lesions were exposed on the surface of the right frontal lobe (**Figure 2(A)**). We administered fluorescein sodium to discriminate the tumor from the normal brain tissue by the previously reported method [2]. The tumor stained deep yellow and was sharply circumscribed. We first separated the tumor from the Rolandic vein and precentral gyrus. As we proceeded into the deep white matter, however, the tumor's borders became more ill-defined. We encountered active bleeding as we removed the rear half of the tumor, the source of which proved to be a dilated vein penetrating the tumor from the AVM (**Figure 2(B)**). As we found it difficult to control the bleeding, we focused on removing the AVM. The nidus was sharply dissected from the cortex, and the feeders were coagulated and cut (**Figures 2(C)** and **(D)**). At the final stage, the draining veins including Rolandic vein, frontal ascending vein and deep veins were treated. The vein within the tumor then altered in diameter to a much more normal size (**Figure 2(E)**). The AVM was completely and safely resected. The resection of the AVM provided palliation of bleeding from the tumor.

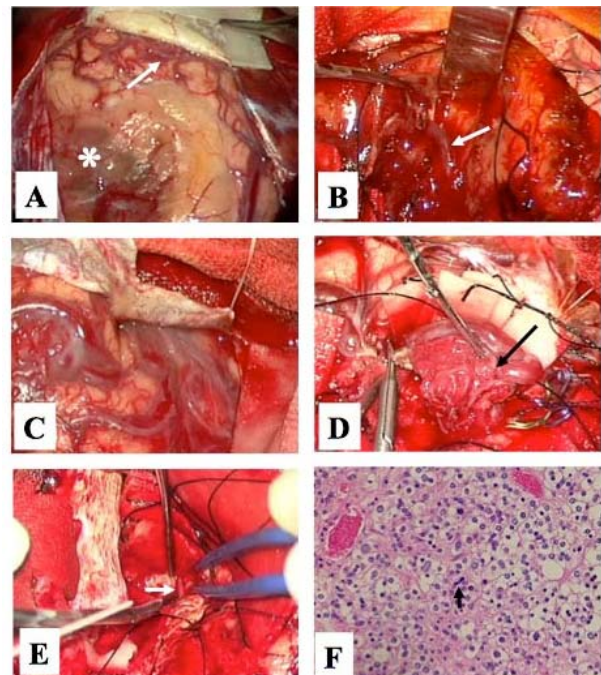


Figure 2. (A)-(E) Intraoperative photographs. (A) The exposed tumor in the right frontal lobe (asterisks) and the AVM (arrow). (B) The dilated red vein penetrating the tumor from the AVM. (C) The cortical red vein of the AVM. (D) The nidus (arrow) was sharply dissected and the final draining vein was cut. (E) Intra-tumoral vein alerted the diameter to the usual (arrow). Intra-tumoral bleeding became controllable. (F) Photomicrograph of hematoxylin & eosin stained slide showing honeycomb appearance with mild nuclear atypia and a few mitoses (arrow), which suggests anaplastic oligodendroglioma ($\times 400$).

The above-mentioned venous sac shriveled. We completed removal of the anterior half of the tumor. The tumor was completely removed using fluorescent navigation. The intra-operative motor evoked potential examination also presented a good response of her forelimb after removal of the tumor. Postoperatively, the patient's level of consciousness was clear and she had no neurological deficits.

3. PATHOLOGICAL EXAMINATION

Formalin-fixed, paraffin-embedded tissue sections were used for hematoxylin-eosin (H & E) staining and immunohistochemistry. H & E staining revealed a sheet-like honeycomb appearance with high cellularity, necrotic foci and nuclear atypia. Mitoses were frequently observed (**Figure 2(F)**). MIB-1 labeling index was 25%. These findings led to a diagnosis as anaplastic oligodendroglioma. The vascular lesion was diagnosed as an AVM, and demonstrated numerous dilated vessels of various sizes.

Following surgery, the patient received external beam radiation therapy with concomitant Temozolomide (TMZ: 75 mg/kg/day). TMZ, however, had to be discontinued

due to pancytopenia a few weeks later. Sixty gray of radiation therapy produced a satisfactory result. Both lesions completely disappeared and there was no recurrence of the tumor 5 year after completion of therapy.

4. DISCUSSION

There have been 74 reported cases of brain tumors associated with AVM to the best of our knowledge (**Table 1**) [3-40]. The rate of this rare association was reported to be 0.1% [19]. There is a male predominance (male: 47 cases, female: 27 cases). The ages ranged from 0 to 70 years of age; the median age was 28.5. The age of onset was unlikely to be related to the association of brain tumor and AVM, and seemed more likely to depend on the kind of brain tumor involved. There are several hypotheses about the etiology and timing of the development of the two lesions in terms of which occurs first and whether one lesion causes the other [26]. Environmental factors and viruses have also been speculated as triggers [26].

Concerning the histology of the brain tumors involved in these cases, there were 12 cases of pilocytic astrocytoma [27,40], 10 of astrocytoma [3,6,8,15,29,30,33,35,36], 6 of malignant astrocytoma [13,18,25,34,36], 20 of oligodendroglioma [4,9,10,20,24,27,38], 3 of glioblastoma [7,19,36], 8 of meningioma [10,11,16,17,21,22,26,32], 5 of acoustic tumors [12], and 3 of pleomorphic xanthoastrocytoma [28,31,39]. Other tumors found in asso-

ciation with AVM include hemangioblastoma [5], hemangiopericytoma [22], ganglioneuroma, [23] subependymal giant astrocytomas [29], and craniopharyngioma [14]. In the remaining 2 cases, histology was simply described as glioma [27,37]. In the present case, the histology was that of an anaplastic oligodendroglioma, which is the type of tumor most frequently associated with AVM.

Concerning the positional relation between the tumor and the AVM, the two lesions were considered “separate” in 18 cases, “intermixed” in 39 cases and “adjacent” in 17 cases (**Table 1**). In our case, it appeared that the tumor had occurred in contiguity with a congenital AVM in the nearby region of the same lobe. Accordingly, the relationship was judged as “separate”. There have been only a handful of cases in which both the AVM and the brain tumor were preoperatively diagnosed using angiography and MRI [29,31-34]. In some reported cases with intermixed AVM in the brain tumor, vascular components were pathologically diagnosed after the surgery. Those were diagnosed as “angioglioma”. In such a case, extensive bleeding during surgery would be assumed; however, the reports make little mention of the operative approach. In the present case, we preoperatively diagnosed both lesions, and decided to remove the tumor first, considering it to be the etiology of the patient’s headaches because we thought the AVM was asymptomatic. As both lesions were located in the right frontal lobe, we could

Table 1. Summary of the positional relationship between brain tumor and AVM.

Tumor histology	Intermixed	Adjacent	Separate	References
Pilocytic astrocytoma	11	1	0	[27,40]
Astrocytoma	2	5	3	[3,6,8,15,29,30,33,35,36]
Malignant astrocytoma	2	2	2	[13,18,25,34,36]
Glioblastoma	1	0	2	[7,19,36]
Oligodendroglioma	14	2	4	[4,9,10,20,24,27,38]
Meningioma	1	4	3	[10,11,16,17,21,22,26,32]
Acoustic tumor	5	0	0	[12]
Hemangioblastoma	1	0	0	[5]
Hemangiopericytoma	0	1	0	[22]
Craniopharyngioma	0	0	1	[14]
Ganglioneuroma	0	1	0	[23]
PXA	1	0	2	[28,31,39]
SEGA*	1	0	0	[29]
Glioma	0	1	1	[27,37]
Total	39	17	18	

*SEGA: subependymal giant astrocytoma.

fortunately observe them in the same bone window at the operation. The tumor was sharply dissected from the Rolandic vein at the beginning of the operation; however, the tumor's boundaries were less clear in the deep white matter. We had difficulty controlling bleeding from a dilated vein passing through the tumor from the AVM to the superior sagittal sinus. The vein was one of the draining veins of the AVM. Although the preoperative angiogram had not revealed them, we should have taken them into consideration from the T2 WI that showed some flow void signs passing through the tumor. We proceeded to remove the AVM with a general procedure. Fortunately, we could safely remove the nidus from the superficial frontal cortex. An intraoperative angiogram showed no residual nidus. It followed that we could dramatically diminish the bleeding and completely remove the tumor.

There have been 5 case reports with the separate type [11,20,21,24,28], in which both of the lesions were removed. Two cases had a good clinical course as the present case [24,28]. As a whole, in 51 cases described the clinical results, 23 cases were good recovery, 18 were moderate disability and 10 cases were dead. Accordingly, the entity does not necessary, which have a good prognosis.

In conclusion, in a case of brain tumor nearby associated with AVM, a large craniotomy including both lesions should be planned to remove the nidus, allowing one to control bleeding from the AVM even if it was preoperatively asymptomatic. The procedures will make a good clinical course.

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LIST OF ABBREVIATIONS

AVM: arteriovenous malformation

MRI: magnetic resonance image

WI: weighted images

Gd-DTPA: gadolinium-diethylenetriamine penta-acetic acid

PET: positron emission tomography

H&E: hematoxylin-eosin;

TMZ: temozolomide