Ruptured Tectal Arteriovenous Malformation Demonstrated Angiographically After Removal of an Unruptured Occipital Lobe Arteriovenous Malformation
—Case Report—

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Abstract
We report a case of ruptured tectal arteriovenous malformation (AVM) that was demonstrated angiographically only after removal of an unruptured occipital AVM. A 57-year-old man presented with sudden onset of diplopia and tinnitus. Computed tomography revealed a small hemorrhage in the right tectum mesencephali with intraventricular hemorrhage. Magnetic resonance imaging and angiography disclosed AVM in the right occipital lobe which was separate from the hemorrhagic lesion. Angiography demonstrated that the right occipital AVM was fed by the parieto-occipital artery and drained into the superior sagittal sinus and vein of Galen. However, no abnormal vascular lesion was detected near the tectum mesencephali. As venous hypertension was considered the reason for hemorrhage, the occipital AVM was completely resected. Postoperative angiography demonstrated disappearance of the occipital AVM, but it also disclosed a small tectal AVM fed by branches from the superior cerebellar artery, which had not been detected on preoperative angiography. This was considered the true cause of hemorrhage, and gamma knife surgery was accordingly performed. Even if an AVM is demonstrated, if the lesion does not correspond to the hemorrhage we recommend serial angiographical evaluation so that a small AVM is not missed.

Key words: multiple arteriovenous malformations, angiography, hemodynamics

Introduction
Multiple arteriovenous malformations (AVMs) are relatively rare and account for 0.3% to 3.2% of all AVMs.5) Cases have reported in patients ranging in age from 9 to 49 years, with a mean of 30.6 years.5) Eighty-five percent of multiple AVMs are located supratentorially, the remainder are both supra- and infra-tentorial.5) Multiple AVMs differ somewhat from common solitary AVMs in terms of clinical course and surgical strategy, because of the influence of hemodynamic changes during resection of multiple lesions.6,7) We present a case of multiple AVMs in which initial angiography revealed only solitary AVM; however, after removal of the first AVM, repeat angiography disclosed another AVM that was the true cause of hemorrhage. To the best of our knowledge, this is the first such case to be reported, and this situation would appear extremely rare. Nonetheless, clinicians should consider multiple AVMs as a possible cause of hemorrhage that appears unrelated to solitary AVM.

Case Report
A 57-year-old man presented with sudden onset of diplopia and tinnitus. On admission, neurological examination revealed Parinaud’s sign, right trochlear nerve palsy, left sensorineural hearing disturbance, and left-sided hypesthesia including left face. Computed tomography revealed a small hemorrhage in the right tectum mesencephali with intraventricular hemorrhage (Fig. 1). T2-weighted magnetic resonance imaging and angiography disclosed an AVM in the right occipital lobe; this was located some distance from the hemorrhagic lesion (Fig. 2). Angiography demonstrated that the right occipital
Fig. 1 Computed tomography scan revealing a small hemorrhage in the right tectum mesencephali with intraventricular hemorrhage.

Fig. 2 T2-weighted magnetic resonance image (A) and angiogram (B) demonstrating a nidus in the right occipital lobe distant from the hemorrhagic lesion.

Fig. 3 Preoperative left vertebral angiograms, anteroposterior (A) and lateral (B) views, showing a nidus fed by branches of the parieto-occipital artery and draining into the superior sagittal sinus and vein of Galen. Arrows indicate where the ruptured arteriovenous malformation appeared after surgery (A).

Fig. 4 Postoperative left vertebral angiograms, anteroposterior (A) and lateral (B) views, showing a small arteriovenous malformation (arrows) fed by branches of the superior cerebellar artery around the tectum mesencephali which was not detected by preoperative angiography, as well as early venous filling of the vein of Galen (arrowheads).

AVM was fed by two branches of parieto-occipital artery and drained into superior sagittal sinus and vein of Galen (Fig. 3). However, no abnormal vascular lesion was detected in the region of the tectum mesencephali. We speculated that the hemorrhage in the tectum mesencephali could have been precipitated by venous hypertension due to shunting vessels from the occipital AVM. Therefore, the right occipital lobe AVM, classified as Spetzler and Martin grade III, was resected via the occipital interhemispheric approach. Postoperative angiography demonstrated disappearance of the AVM in the right occipital lobe but revealed a small AVM fed by branches from superior cerebellar artery in the region of the tectum mesencephali (Fig. 4). This AVM had not been detected on preoperative angiography. As this AVM was considered to have ruptured, gamma knife surgery was performed with a marginal dose of 15 Gy. The postoperative course was uneventful. Follow-up angiography after 12 months revealed obliteration of the small AVM in the tectum mesencephali and reduction of early venous filling of the vein of Galen (Fig. 5).

Discussion

Multiple AVMs are relatively rare and account for 0.3% to 3.2% of all AVMs.\textsuperscript{5} Sixty-five percent of those are symptomatic present with hemorrhage.\textsuperscript{5}
In general, AVM that are smaller and deeper are more likely to hemorrhage.\textsuperscript{3} In the present case, the AVM that was responsible for hemorrhage, which was diagnosed after removal of the occipital AVM, had these characteristics. In the context of multiple AVM surgery, secondary hemorrhage has occasionally been noted after the first operation, and hemodynamic changes during the different treatment stages have demonstrated previously undiagnosed vascular malformations.\textsuperscript{2,4,7,8} Utsuki et al. and Tada et al. emphasized such a event,\textsuperscript{6,7} and Salcman et al. and de Sousa et al. recommend total resection of all lesions; first ruptured AVM, and then the remaining lesions.\textsuperscript{1,5} In the present case, angiography demonstrated a small tectal AVM after resection of an AVM in the occipital lobe, so the angiographical appearance of the lesion might have been affected by thrombosis of the shunt just after hemorrhage, by absorption of the hematoma, and by hemodynamic changes after resection of the larger AVM.

When imaging reveals unruptured AVM with distant hemorrhage from an unknown source, we emphasize the possibility of occult AVM and recommend a careful preoperative search for such AVM on angiography and prompt follow-up angiography to examine the reorganization of cerebral blood flow after resection of an AVM.

References


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