Rapid development of an intranidal aneurysm with perifocal brain edema in an unruptured cerebral arteriovenous malformation

Case report

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The authors present the case of a 22-year-old man with an unruptured arteriovenous malformation (AVM) in which an intranidal aneurysm had grown in the course of 3 months and was complicated by perifocal brain edema. A left parietal AVM was incidentally diagnosed on magnetic resonance (MR) imaging. No aneurysms were noted on cerebral angiograms obtained simultaneously. Three months later, T₂-weighted MR imaging revealed perifocal brain edema (increased signal intensity in the brain parenchyma adjacent to the nidus). An aneurysm-like signal void was demonstrated in the center of the high-signal area, but no previous hemorrhages could be detected. Angiographic studies revealed an intranidal aneurysm 4 mm in diameter projecting anterolaterally from the nidus. Surgical removal was performed without incident, and no neurological deficits or postoperative complications were observed. An MR image obtained 2 weeks post-surgery revealed complete resolution of the perifocal brain edema. To the authors’ knowledge, this is the first reported case of an unruptured AVM in which an intranidal aneurysm with perifocal brain edema developed rapidly (within a few months).

Key Words • arteriovenous malformation • aneurysm

Intracranial bleeding is the biggest threat in patients with AVMs. Intranidal aneurysm is recognized as one of the major risk factors for such bleeding, and its presence may influence treatment strategy. For incidental small AVMs with no neurological deficit, radiosurgery can be a powerful treatment option. For AVMs with intranidal aneurysms, on the other hand, the possibility of more aggressive treatment should be considered, including surgery, given the high risk of intracranial bleeding during the latent period after radiosurgery.

We report on a patient with an AVM in whom perifocal brain edema developed adjacent to the nidus due to rapid development of an intranidal aneurysm within a period of 3 months.

Case Report

History and Examination. In April 2001, this 22-year-old man with no history of intracranial bleeding or seizure was incidentally found to have an abnormal vascular signal void in the left parietal lobe on MR imaging (Fig. 1). On T₂-weighted MR images no high-intensity area was revealed around the signal void. Angiographic studies demonstrated an AVM with a major 25-mm axis, which was mainly fed by the left pericallosal artery and drained into both the left parietal ascending vein and the internal cerebral vein (Fig.

Fig. 1. Axial T₂-weighted MR images of the middle (left) and upper (right) levels of the nidus obtained at diagnosis, demonstrating no high-signal area in any slice.

Abbreviations used in this paper: AVM = arteriovenous malformation; GKS = gamma knife surgery; MR = magnetic resonance; VEGF = vascular endothelial growth factor.
2); no aneurysm was detected. Because the anterior portion of the nidus was adjacent to the corona radiata, surgical treatment was not considered initially due to a high risk of impending neurological morbidity. Therefore, GKS was recommended as a treatment modality.

The patient was admitted to our hospital in July 2001 to undergo GKS. Stereotactic T₂-weighted MR imaging revealed increased signal intensity in the brain parenchyma adjacent to the nidus. An aneurysm-like signal void was demonstrated in the center of the high-signal area (Fig. 3). No previous bleeding could be detected. Stereotactic angiograms obtained after MR imaging revealed an intranidal aneurysm with a 4-mm diameter, which was filled in the early arterial phase before substantial venous filling and projected anterolaterally from the nidus (Fig. 4). No apparent stenosis or occlusion of the draining veins resulting in hemodynamic stress to the nidus was observed. Three-dimensional definition of the intranidal aneurysm attained using the Leksell GammaPlan program (Elekta Instruments AB, Stockholm, Sweden) indicated that the aneurysm seen on the angiogram was coincident with the signal void at the center of the edematous area on MR images (Fig. 5). At this point, because we inferred that risk of bleeding was very high, we decided to discontinue GKS and planned to perform conventional surgery.

Operation and Postoperative Course. Removal of the nidus was achieved with no resulting neurological deficit. Intraoperative findings revealed an intranidal aneurysm with a very thin vascular wall arising from the nidus. No evidence of hemorrhage from the nidus was observed. Complete removal of the nidus was confirmed on postoperative angiography. Magnetic resonance images obtained 2 weeks postsurgery revealed complete resolution of the perifocal brain edema adjacent to the nidus (Fig. 6).

Discussion

To our knowledge, this is the first report of the rapid development of an intranidal aneurysm associated with
marked perifocal brain edema. Although it is known that a pseudoaneurysm that develops after bleeding of an AVM sometimes grows very rapidly, there was no sign of bleeding on either MR imaging or intraoperative examination in our case.

The reason the aneurysm developed so rapidly in this case is not clear. One possible explanation is that a hemodynamic change within the nidus increased stress to the wall of the microaneurysm, which was occult during the initial angiographic study. It has been reported that occlusion or severe stenosis of the major draining vein may lead to formation of perifocal brain edema and neurological deterioration. Moreover, Meng and Okada found histological evidence of microscopic occlusion of small draining veins within a nidus surrounded by loose connective tissue. This type of stenosis may raise the intranidal pressure, resulting in formation of an intranidal aneurysm. Although we did not observe changes in the draining system of the AVM between two angiographic examinations, the aforementioned

Fig. 3. Axial T2-weighted MR images of the middle (left) and the upper (right) levels of the nidus obtained 3 months after diagnosis. An area with increased intensity is observed in the brain parenchyma adjacent to the nidus. An aneurysm-like signal void is demonstrated in the center of the perifocal brain edema (arrow).

Fig. 4. Anterior (A and C) and lateral (B and D) views on left ICA angiograms obtained 3 months after diagnosis. An intranidal aneurysm (arrows) is demonstrated in the upper anterolateral portion of the nidus (A and B). No stenosis or occlusion of the draining veins is observed (C and D).
mechanism may explain rapid development of an intranidal aneurysm in a very limited part of the nidus, as was observed in our case.

Although it has been reported that giant aneurysms cause extensive brain edema due to mass effect,\(^3\) the aneurysm in our patient appeared to be too small to induce obvious brain edema. Another possible explanation is that VEGF, one of the growth factors that regulates angiogenesis, may also play an important role in the formation of intranidal aneurysms. In an earlier report,\(^10\) VEGF was found to be expressed strongly within the wall of aneurysms in some cases and was said to play some role in the formation and development of aneurysms; VEGF is also known to promote vascular permeability.\(^4\) Although we did not check its concentration pre- or postoperatively, we suspect that VEGF or other angiogenic factors may play roles in the rapid development of intranidal aneurysms with perifocal brain edema, as occurred in our case.

Intracranial bleeding is the biggest threat in patients with AVMs. In recent studies it has been shown that the annual bleeding rate of AVMs accompanied by intranidal aneurysms is as high as 8.5 to 9.8%.\(^3\) It can thus be speculated that the risk of bleeding is much higher when the aneurysm is growing rapidly. As in our case, because the risk of bleeding is considered to be very high, instead of GKS, early

\[\text{FIG. 5. Angiograms of the left ICA (A) and serial axial T}_{2}\text{-weighted MR images (B) displayed on workstations running the Leksell GammaPlan program. A: Contours of the intranidal aneurysm on anterior (left) and lateral (right) views are indicated by white circles. B: The three-dimensional location of the aneurysm on the angiograms is superimposed onto the serial axial T}_{2}\text{-weighted MR images. A quadrilateral (arrow), which is formed by the intersections of four straight lines, indicates the position of the aneurysm on angiograms. The aneurysm is coincident with the signal void at the center of the edematous area on MR images.}\]

\[\text{FIG. 6. Axial T}_{2}\text{-weighted MR images obtained 2 weeks after surgical removal of the AVM. Increased signal (edema) was completely resolved postoperatively both in the middle (left) and upper (right) levels of the nidus.}\]
surgical removal of the entire malformation is the most effective method of treating an AVM in which rapid development of an intranidal aneurysm is exhibited.

Conclusions

When an AVM with rapid development of perifocal brain edema around the nidus is noted on follow-up MR images, angiographic examination followed by prompt surgical treatment is indicated because of the possibility of a newly developed intranidal aneurysm.

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References


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