Risk of Rebleeding in Arteriovenous Malformations Due to Impaired Venous Drainage After Radiosurgery

—Case Report—

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Abstract

A 27-year-old woman presented with rebleeding from an intracranial arteriovenous malformation (AVM) 6 years after radiosurgery. Cerebral angiography demonstrated venous drainage change into a single drainer and cortical reflux due to drainage occlusion into the superior sagittal sinus. During surgery, multiple small feeders thought to be occluded on preoperative angiography caused brain swelling, hindering resection of the AVM border plane. Flow changes after radiosurgery, especially impaired venous drainage, may have increased the tendency to rebleeding of the AVM. Scheduled angiography after radiosurgery is recommended, and retreatment for residual AVMs is preferable, especially if venous drainage change occurs.

Key words: intracranial arteriovenous malformation, radiosurgery, venous drainage, rebleeding, cortical reflux

Introduction

Intracranial arteriovenous malformations (AVMs) are relatively uncommon and predominantly occur in young adults. More than 50% of AVMs result in intracranial hemorrhage, which can cause serious neurological symptoms or death.3,11) Radiosurgery is recommended for patients with small lesions (volumes <10 cm³ or maximum diameter <3 cm), especially those located in eloquent or surgically inaccessible areas.11) Complete AVM obliteration occurs in 80% of patients within 2–3 years. Radiosurgery decreases the risk of hemorrhage in patients with AVM, even before angiographic evidence of obliteration.7) However, obliteration takes years and the risk of
hemorrhage remains at 1.8–5% per year. The timing and options of retreatment for residual AVMs remain largely unknown. We describe a case of direct surgery for a rebleeding AVM with venous drainage change after radiosurgery.

Case Report

A 27-year-old woman initially presented to another institution with headache caused by intraventricular hemorrhage. Cerebral angiography showed a right parietal AVM (Spetzler-Martin grade III) that measured 1.4 × 2.2 × 3.2 cm. The AVM was fed by anterior cerebral artery (ACA) and middle cerebral artery (MCA) branches with several draining veins into the superior sagittal sinus (SSS) (Figs. 1A and 2A, D). The AVM nidus volume was 5.2 cm³. The patient received gamma knife radiosurgery with 18 Gy at the 50% isodose (36 Gy maximum). Angiography performed 2 years later showed a partially obliterated AVM, with prominent disappearance of the medial part of the nidus (Fig. 1B). A new drainer arose from the lateral part with anterograde venous drainage (Fig. 2B, E).

Six years after radiosurgery, the patient first presented to us with altered mentation (Glasgow Coma Scale score 13: E3, V4, M6), left hemiplegia, and paresthesia caused by right parietal subcortical hemorrhage. Cerebral angiography demonstrated an incompletely obliterated AVM located in the postcentral gyrus, caudal to the hematoma, with mainly ACA branch feeders (superior internal cerebral artery), and an occluded main drainer at the entry point to SSS, leading to superficial sylvian and Labbe veins as cortical reflux without SSS occlusion (Figs. 1C and 2C, F).

Emergency surgery was performed. Two arterial feeders from the ACA were first coagulated, followed by attempted excision of the nidus. However, the brain swelling worsened. The lateral border plane was rapidly resected and bleeding from multiple small feeders from MCA was managed. None of these sites were detected by preoperative cerebral angiography. Finally, total removal was accomplished. Postoperative computed tomography and angiography confirmed complete removal of the AVM (Fig. 1D). Histological examination showed the AVM consisted of clusters of abnormal arteries and arterialized veins with irregular vessel wall structure, and an intervening capillary bed, mixed with hyalinization. The patient made an excellent recovery and lived independently with mild left hemiparesis at 2-year follow up.

Discussion

The configuration of the venous drainage system may be predictive of bleeding of AVMs. Although venous stenosis or a single drainer are not independent predictors of AVM rupture in some studies, venous drainage impairment was found to be positively correlated on theoretical grounds. A single draining vein, impaired venous drainage, and deep venous drainage may be associated with increased risk of bleeding. Venous drainage changes are often observed after radiosurgery of AVMs, as newly developed 'en passant' feeding arteries or stenosis of the main venous drainage. Hemorrhagic risk after radiosurgery is correlated with morphological parameters including venous hypertension, deep venous drainage, AVM-associated aneurysms, periventricular location, in-
complete coverage, and minimum dose.\textsuperscript{6,10}

In our case, we confirmed that change of the draining system into single venous drainage and cortical reflux occurred due to occlusion of several drains into the SSS. Irradiation may directly compromise venous outflow or indirectly promote venous thrombosis resulting from altered intranidal hemodynamics. Impaired venous drainage caused by drainer occlusion may only raise nidus vessel pressure, and convert AVMs prone to rebleeding through venous drainage changes. In addition, a new drainer occurred from the residual nidus. The change of flow dynamics may also have contributed to the bleeding tendency of the incompletely obliterated AVM.

Incomplete AVM radiosurgery was associated with targeting error, low treatment doses (in particular < 15 Gy), large AVM volume, and high Spetzler-Martin grade.\textsuperscript{21} This AVM may have represented a challenge for radiosurgery as a definitive treatment. The radiosurgery-based grading score used was $0.1 \times 5.2 \text{ (volume)} + 0.02 \times 27 \text{ (age)} + 1 \text{ (parietal location)} = 2.6$, which corresponds to less than 60% probability of an excellent outcome.\textsuperscript{12} The relatively large volume of the lesion resulted in application of a peripheral irradiation dose of only 18 Gy, which may have contributed to the incomplete obliteration. Furthermore, the occluded main drainer was included in the irradiated field, and direct irradiation of the drainers might have led to the complication. Thus, accurate exposure focused on the nidus and feeders of AVM is critical, with sparing of the sinus or main draining.

Radiosurgical retreatment is considered safe and effective in cases of failed radiosurgery for AVM.\textsuperscript{2} Direct surgery was also recommended for remnant AVMs at 3 years after radiosurgery, even in asymptomatic patients, because radiation-induced changes in AVMs could facilitate microsurgical resection.\textsuperscript{13} In contrast, microsurgical resection may be challenging because of the difficult preparation of firm perinodal tissue and surrounding deep feeding arteries due to neoangiogenesis.\textsuperscript{11} In our case, we had difficulty with multiple feeders that we thought were occluded. The use of the correct procedure during direct surgery to remove the nidus is critical to improve outcome, and careful examination for missed small feeders is important, particularly if no prior radiosurgery has been performed.

In summary, incomplete obliteration of radiated AVMs remains a risk factor for rebleeding. Scheduled cerebral angiography after radiosurgery is recommended to evaluate the morphological changes of irradiated AVMs. Retreatment for residual AVMs is preferable to prevent rebleeding. We suggest that flow changes after radiosurgery, especially impaired venous drainage by drainer occlusion, may cause cerebral hemorrhage, and that immediate radiosurgical retreatment or direct surgery is required if venous drainage change is observed.

References


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