Chronic encapsulated expanding hematoma in association with gamma knife stereotactic radiosurgery for a cerebral arteriovenous malformation

Case report

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The authors report a rare case of a patient with a chronic encapsulated expanding hematoma and progressive neurological deterioration who presented 2 years after gamma knife radiosurgery for a cerebral arteriovenous malformation (AVM). A tough capsule containing multiple layers of organized hematoma resulting from previous bleeding was confirmed surgically. Histological examination revealed that the capsule consisted of a dense collagenous outer layer and a granulomatous newly vascularized inner layer with marked fibrosis. Hemosiderin deposits were frequently observed in the inner layer, which suggested recurrent minor bleeding from fragile vessels in this layer. An AVM was found in the hematoma, which had degenerated as the result of radiosurgery. A cross-section of the abnormal vessels showed various stages of obliteration due to intimal hypertrophy. The clinical course, radiological features, and histological findings in this case were compatible with those of previously reported chronic encapsulated hematomas. A possible mechanism of hematoma formation and its expansion are discussed.

KEY WORDS • chronic encapsulated hematoma • arteriovenous malformation • gamma knife • radiosurgery

During the last two decades, stereotactic radiosurgery has become a popular therapeutic alternative for the treatment of cerebral arteriovenous malformations (AVMs). More than 6000 patients with AVMs have undergone this procedure. For small AVMs, single-fraction, high-dose, small-field focused radiation therapy has been demonstrated to be an effective, noninvasive method for nidus obliteration. However, complications have been reported in the use of this procedure, including adverse reactions to radiation and hemorrhage from the treated but not yet obliterated AVMs.

In the present report, we describe a case of encapsulated intracerebral hematoma that developed after radiosurgery during the course of obliterating an AVM. The AVM, which had caused an ordinary bleeding episode prior to radiosurgery, was altered by the radiosurgery, causing hemorrhages that led to the formation of an encapsulated intracerebral hematoma. To our knowledge, this is the first report of this type of rebleeding from a residual AVM after radiosurgery.

The etiology of the chronic encapsulated intracerebral hematoma is poorly understood. Our experience adds to the knowledge concerning pathogenesis of this peculiar hematoma.

Case Report

This 19-year-old left-handed man presented with a sudden onset of left-sided hemiparesis associated with nausea and vomiting in February 1991. He was referred to a local hospital. Computerized tomography (CT) scanning of the head showed intracerebral bleeding in the right basal ganglia (Fig. 1). Cerebral angiography revealed an AVM with a maximum diameter of 2.5 cm. After initial conservative management, he was transferred to our hospital for radiosurgical treatment.

Radiosurgery. Radiosurgery using the 201-source 60Co gamma knife was performed in April 1991. While the patient was receiving a local anesthetic, the Leksell model “G” stereotactic coordinate frame (Elekta Instruments, Atlanta, GA) was fixed to his skull. Complex isodose planning on the basis of the stereotactic cerebral angiography was performed. The AVM was located in the right internal capsule and was fed mainly by the lenticulostriate arteries, which drained into the basal vein. Twenty Gray of radiation was delivered at the periphery of the AVM in a single dose using six overlapping 14-mm collimator fields (Fig. 2).

Clinical Course. The patient’s spastic hemiparesis improved gradually after treatment. At his 6-month follow-
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up review, contrast-enhanced CT scanning showed no ni-
dus enhancement (Fig. 3b; compare with Fig. 3a); howev-
er, cerebral angiography revealed the presence of residual
AVM (Fig. 3c and d).
In February 1992, 10 months after radiosurgery, the
patient suffered further deterioration of his hemiparesis.
Magnetic resonance (MR) imaging with gadolinium di-
ethylenetriamine pentaacetic acid was performed, which
showed a localized gadolinium enhanced lesion on the
right internal capsule (Fig. 4 left). Initially, the lesion was

Fig. 1. Computerized tomography scan showing initial intra-
cerebral hemorrhage in the right basal ganglia.

Fig. 2. Stereotactic right internal angiograms: anteroposterior (left) and lateral (right) views demonstrating an arterio-
venous malformation in the right internal capsule. The arrowheads indicate the 50% isodose line. At radiosurgery, 20 Gy
was administered to the 50% isodose line using six isocenters of radiation with the 14-mm collimator (maximum dose
40 Gy).

Fig. 3. a and b: Computerized tomography scans with contrast enhancement. a: Preradiosurgery scan showing contrast enhance-
ment of the nidus on the right internal capsule (arrows). b: Six-
month follow-up scan showing disappearance of nidus enhance-
ment. c and d: Right internal angiograms obtained 6 months after
radiosurgery: anteroposterior (c) and lateral (d) views demonstrat-
ing major, but incomplete, obliteration of the arteriovenous
malformation nidus. Although the draining vein has not persisted,
the nidus is still detected as a small stained area.
diagnosed as radiation necrosis and was treated conservatively. However, in September 1992, follow-up MR imaging revealed enlargement of the hyperintense area on a T2-weighted image (Fig. 4 right). In March 1993, 2 years after radiosurgery, the patient developed severe headache, nausea, and vomiting. A CT scan demonstrated a large heterogeneous high-density mass with perifocal edema in the right basal ganglia (Fig. 5 left). A decompressive right frontotemporal craniectomy with hematoma evacuation was performed.

First Operation. The yellow-colored ependyma above the thalamus was incised via an interhemispheric transventricular approach, and the hematoma cavity was entered. The hematoma showed various stages of organization with granulation tissue, which suggested minor repetitive bleeding. A granulomatous and partially thrombosed nidus was found in the hematoma and was removed in part.

Postoperative Course. Subsequently, the patient developed normal-pressure hydrocephalus, and a ventriculoperitoneal shunt was placed. He was discharged in December 1993 with additional visual impairment due to sustained intracranial hypertension. He remained stable with no further neurological deterioration until September 1994, when a follow-up CT scan again demonstrated an expansion of the ring-enhancing encapsulated hematoma into the right lateral ventricle (Fig. 5 center and right). The patient’s family refused further surgical treatment of the lesion. He was treated conservatively until November 1994, when he gradually developed a disturbance of consciousness. The patient was readmitted to our hospital in a semicomatose state and an emergency operation was performed.

Second Operation. The hematoma was exposed via a right frontotranscortical approach. The thick elastic capsule was located 2.5 cm below the cortex. Many small abnormal vessels entering the capsule were found to be coagulated. The organized hematoma, partially thrombosed nidus of the AVM, and hematoma capsule were excised totally. Following surgery, the patient’s consciousness improved. No further progression has been observed in his motor and visual dysfunction. He was discharged from the hospital 6 weeks after the second surgery.

Histological Examination. The hematoma contained organized clots at various stages of development. The wall consisted of an outer layer of dense collagenous tissue and an inner layer of “angiomatoid” fibroblastic granulation tissue with numerous dilated capillaries. Hemosiderin deposits were frequently seen in this inner layer (Fig. 6 left). There were medium-sized AVM vessels showing various stages of obliteration due to intimal hypertrophy with and without fragmentation of the internal elastic layer. Exudation was also seen in the nidus (Fig. 6 right). Small fragments of brain tissue surrounding the capsule showed reactive gliosis, but inflammatory and neoplastic changes were not observed.
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**Discussion**

Radiosurgery is believed to cause obliteration of the AVM nidus by inducing cell proliferation of the vascular lumen and subsequent wall thickening. Complete angiographic obliteration of an AVM is the goal of radiosurgery; no bleeding from an AVM that has been completely occluded angiographically has been reported following the treatment. Before angiographic cure is attained, there remains a risk of further bleeding from the incompletely or partially obliterated AVMs. Indeed, Guo, et al., reported a case of rebleeding from a tiny remnant of AVM 49 months after radiosurgery. We report here an unusual type of hematoma that developed at the site of a partially obliterated AVM following radiosurgery. This hematoma is unique in its gradual clinical onset, capsule formation, and self-perpetuating expansion, which are compatible with the previously reported clinical entity of chronic encapsulated intracerebral hematoma. In our case, the possibility cannot be excluded that continuous minor bleeding from the residual nidus resulted in hematoma expansion. However, histological examination suggested that it was due to repetitive bleeding from the fragile vessels contained in the thick hematoma capsule.

Although there are numerous reports of chronic encapsulated intracerebral hematoma, the mechanism for capsule formation and progressive enlargement still remains uncertain. In our review of the literature, 29 cases of this entity were reported. Vascular anomaly was frequently seen as a bleeding source in those cases, which included five angiographically occult AVMs, six cavernous angiomas, one venous angioma, and one unclassified vascular malformation. No clear causes were established in the remaining cases, except for one case of metastatic adenocarcinoma. Several authors assumed that some chronic encapsulated intracerebral hematomas without clear bleeding sources were related to vascular malformations. Roda, et al., suspected that the cause of such a hematoma was probably small vascular malformations that were destroyed or thrombosed during the hemorrhagic episodes. Pozzati, et al., emphasized that occult “self-destroying” vascular malformation was responsible for the initial hemorrhage. Hirsh, et al., proposed that the hematoma capsule was made of fibroblasts derived from the abnormal vessels of occult vascular malformations. Our case also suggests that subclinical bleeding from the vascular malformation is a trigger for this peculiar hematoma formation. During the process of AVM degeneration after radiosurgery, repetitive minor bleeding around the partially thrombosed AVM and fibroblastic proliferation from the degenerated nidus may form a neovascularized capsule. Then, repeated intracapsular bleeding and exudation may expand the hematoma in a fashion similar to chronic subdural hematomas. Initially, the AVM in our case had caused an ordinary intracerebral hematoma. Radiosurgery changed the character of bleeding from the AVM to form a chronic encapsulated intracerebral hematoma.

The present case suggests that rebleeding during the latency interval between radiosurgery and angiographic cure could occur in a different manner from the original bleeding. The radiological features of this intracerebral hematoma were quite misleading. Initially, we suspected the lesion in our patient was due to radiation necrosis; Statham, et al., reported a case of radiation necrosis following radiosurgery that presented with local mass sign and mimicked a malignant tumor. To manage patients with these symptoms properly, neurosurgeons need to be aware of the clinical behavior and radiological findings of this fairly rare condition associated with radiosurgery for AVMs. We believe surgical extirpation is the only way to treat this type of bleeding.

**References**


Manuscript received March 21, 1995. Accepted in final form October 24, 1995.
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