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Successful Endoscopic Resection of a Chronic Expanding Hematoma Following Gamma Knife Surgery for Cerebral Arteriovenous Malformation: A Case Report

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Abstract

Gamma knife surgery is useful for treating cerebral arteriovenous malformations. However, some radiation-induced long-term complications have been reported. One of these is a chronic expanding hematoma. We present a case of chronic expanding hematoma, successfully treated with endoscopic resection. The patient, a woman in her 30s, experienced a cerebral hemorrhage 17 years ago associated with an arteriovenous malformation in the corpus callosum. The lesion was completely embolized with n-butyl-2-cyanoacrylate embolization twice, followed 2 years later by gamma knife surgery. The patient presented to the emergency room with headache and left hemiplegia. A computed tomography scan showed hemorrhagic changes consistent with the cystic lesion and worsening edematous changes around it. An endoscopic tumor resection (interhemispheric approach) was performed, and most of the lesion was removed. Pathology did not detect any neoplastic lesions, and a diagnosis of chronic expanding hematoma was performed based on the presence of abnormal vascular neoplasia. The postoperative course was uneventful, and the headache promptly resolved. The pathophysiology of chronic expanding hematoma involves slow and progressive hematoma expansion due to repeated local hemorrhage, causing intense cerebral edema around the lesion. Surgical removal is effective, and edematous changes and neurological symptoms can be quickly relieved after surgery. Endoscopic surgery is particularly effective for deep lesions owing to its ability to manipulate within a narrow surgical field. In this case, the lesion was removed with minimal invasiveness and no complications, leading to early symptom relief and resolution of the surrounding brain edema changes.

Keywords: chronic expanding hematoma, gamma knife surgery, arteriovenous malformation, endoscopic surgery

Introduction

Cerebral arteriovenous malformations (AVMs) are rare, but they have a hemorrhage rate of 2% to 4.5% per year.[1-3] In addition, each hemorrhage is associated with an approximate 18% mortality. A chronic expanding hematoma (CEH) is a late complication induced by stereotactic radiosurgery.[1,4-8] It is a rare condition with few reports in the literature.[1,4,5,8] We report a case of CEH that occurred 17 years after gamma knife surgery (GKS) for an AVM, successfully treated with endoscopic resection.

Case Report

A 32-year-old woman experienced an intracerebral hemorrhage 17 years ago associated with Spetzler-Martin grade II AVM in the corpus callosum (Fig. 1A-D). The lesion was embolized twice with n-butyl-2-cyanoacrylate (NBCA), followed by GKS for the remaining lesion (Fig. 1E). Digital subtraction angiography (DSA) 3 years after GKS showed complete obliteration of the lesion (Fig. 1F). Ten years after GKS, a cystic lesion, previously unseen, appeared around the area of the original AVM (Figs. 1G and 1H). The cyst alternately grew and shrank, and the patient continued to be monitored (Fig. 1I). However, both the lesions

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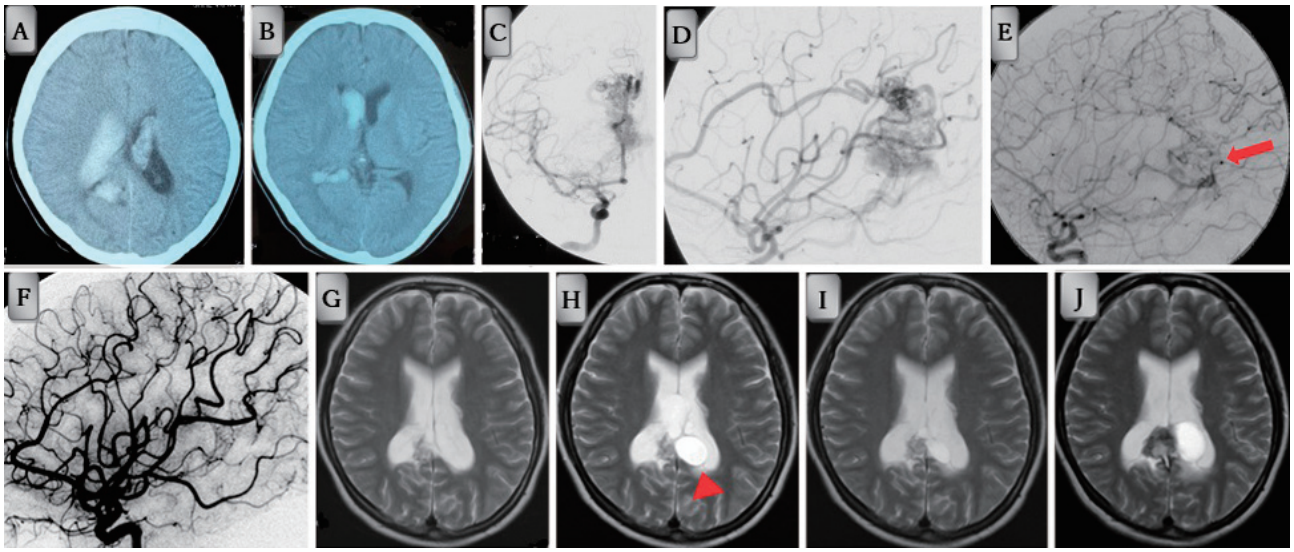

Fig. 1
(A–D): Internal carotid angiograms show the hemorrhagic AVM of the parasplenial 17 years ago.
(E): A residual lesion was observed after 2 embolizations with NBCA (red arrow).
(F): After GKS, the AVM was completely occluded.
(G): Axial T2-weighted MRI obtained 8 years after GKS.
(H): Axial T2-weighted MRI obtained 10 years after GKS. A cystic lesion appeared on the left side of AVM (red arrowhead).
(I): Axial T2-weighted MRI obtained 12 years after GKS. The cyst repeated increase and decrease.
(J): Axial T2-weighted MRI obtained 14 years after GKS. The lesion around AVM increased.
AVM: arteriovenous malformation; GKS: gamma knife surgery; MRI: magnetic resonance imaging; NBCA: n-butyl-2-cyanoacrylate; T2: type 2

and cysts began to increase in size 14 years after GKS (Fig. 1J).

The patient presented to the emergency room with a headache and left hemiparesis 15 years after GKS. A computed tomography scan of the brain showed hemorrhagic changes consistent with the lesion and worsening edematous changes around it (Fig. 2A and B). Moreover, the cystic lesions surrounding the original AVM had disappeared (Fig. 2C and D). DSA showed no recurrence of the AVM (Fig. 2E and F). The patient was transferred to our hospital for treatment. Although dexamethasone administration improved the left hemiparesis, intermittent severe headaches persisted. Owing to increasing edematous changes around the lesion, an endoscopic tumor resection (interhemispheric approach) was performed.

The surgery was performed under general anesthesia with the patient in the right three-quarter prone position. Bilateral internal cerebral veins (ICVs) were identified at depth of surgical field through a posterior interhemispheric approach after right parietal craniotomy across the superior sagittal sinus (SSS) (Fig. 3A and B). A red lesion was observed on the ventral side of the ICVs with a nearby white embolized vessel (Fig. 3C and D). The bilateral lateral ventricles were identified while removing the lesion piece by piece, and the lesion attached to the ventricular wall and choroid plexus was removed (Fig. 3E-I). The lesion on the dorsal side was difficult to remove owing to strong adherence to the ICVs (Fig. 3J and K). Therefore, the remaining lesion was coagulated (Fig. 3L).

Pathological findings indicated organizing tissue on the outside, encasing the internal hematoma like a capsule. Numerous abnormal vessels with large and small dilatations were observed within the hematoma, leading to a pathological diagnosis of CEH (Fig. 4A-D). The postoperative course was uneventful, and the patient experienced rapid relief from headaches after surgery. She was discharged 16 days after the operation. Magnetic resonance images (MRI) of the head showed marked improvement in edematous changes, which continued to shrink (Fig. 4E-J).

Discussion

In AVM treated with stereotactic radiosurgery, CEH is a rare condition; however, the incidence has been increasing in recent years.[4,8] CEH has been suggested to be associated with vascular malformations and is reported in cases involving AVM.[4,5,8] Nakamizo et al.[8] reported that approximately 40% of CEH cases are related to vascular malformations. In addition, Abou et al.[1] found that most CEH cases developed 2 to 12 years after stereotactic radiosurgery.

The pathophysiology of CEH is characterized by a slow onset and progressive expansion of the hematoma due to

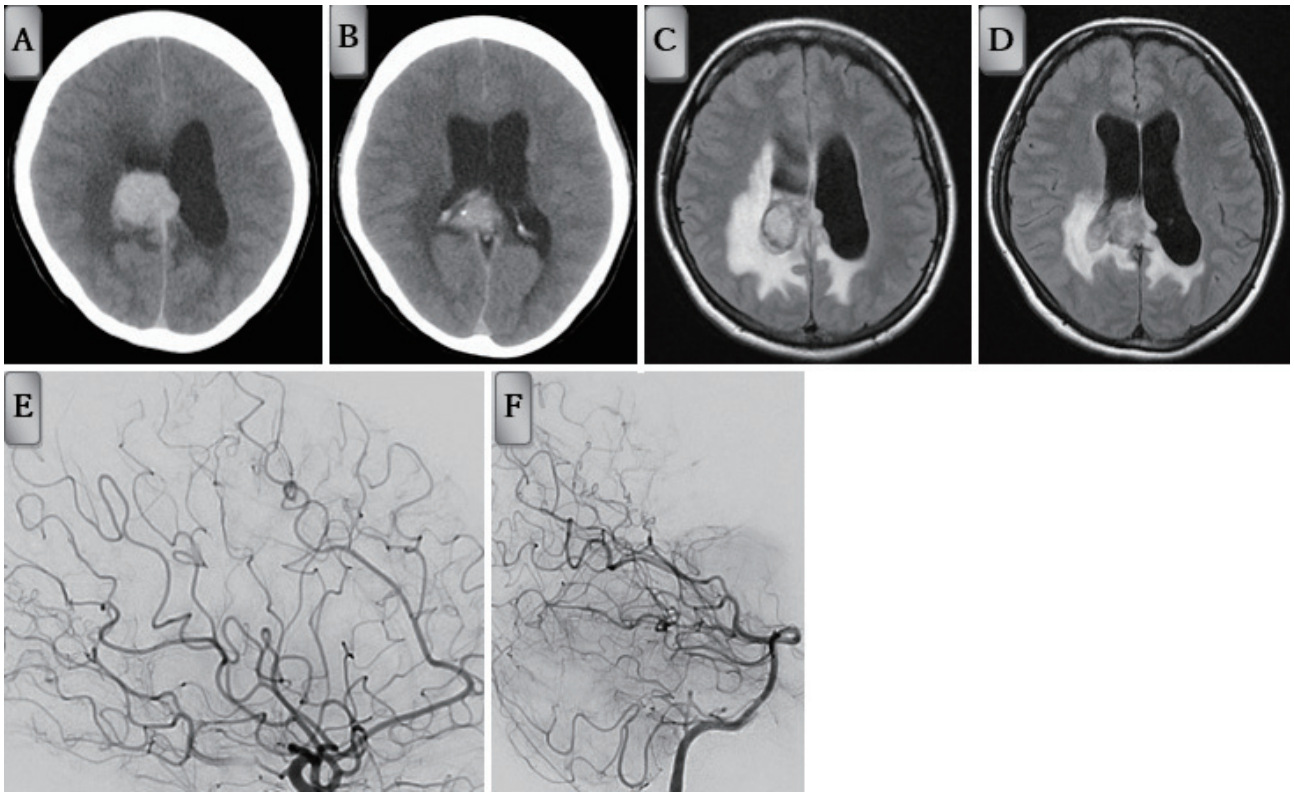

Fig. 2
(A–D): A headache and left upper and lower limb paralysis suddenly developed in the patient, 15 years after GKS. Computed tomography images and FLAIR MRI obtained showed increased lesion size and internal hemorrhagic changes. Edematous changes also appeared around the lesion.
(E, F): Digital subtraction angiography revealed that the AVM had not recurred.
AVM: arteriovenous malformation; FLAIR: fluid-attenuated inversion recovery; MRI: magnetic resonance imaging

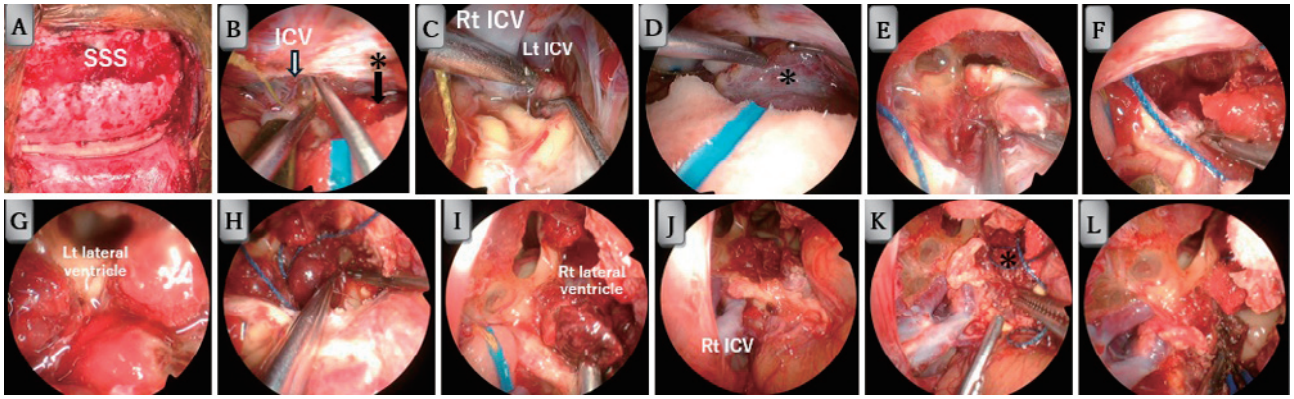

Fig. 3
(A–L): The operation was performed at posterior interhemispheric approach after right parietal craniotomy across the SSS. A red lesion (*) was observed on the ventral side of the ICVs with a nearby white embolized vessel. Most lesions were removed piece by piece. However, the areas of strong adhesion to the bilateral ICVs were left in place, and the remaining lesion was coagulated.
ICV: internal cerebral vein; SSS: superior sagittal sinus

repeated local bleeding.[4] However, the exact mechanism of CEH development remains unclear.[1,4,5,8] Histopathologically, CEH comprises an encapsulated organized hematoma.[4] The inner layer of the capsule exhibits abnormal angiogenesis and proliferation of myofibroblasts, whereas the outer layer comprises fibrous and granulation tissues.[1,4] It is believed that CEH progresses slowly owing to repeated bleeding and exudation from fragile vessels in the inner

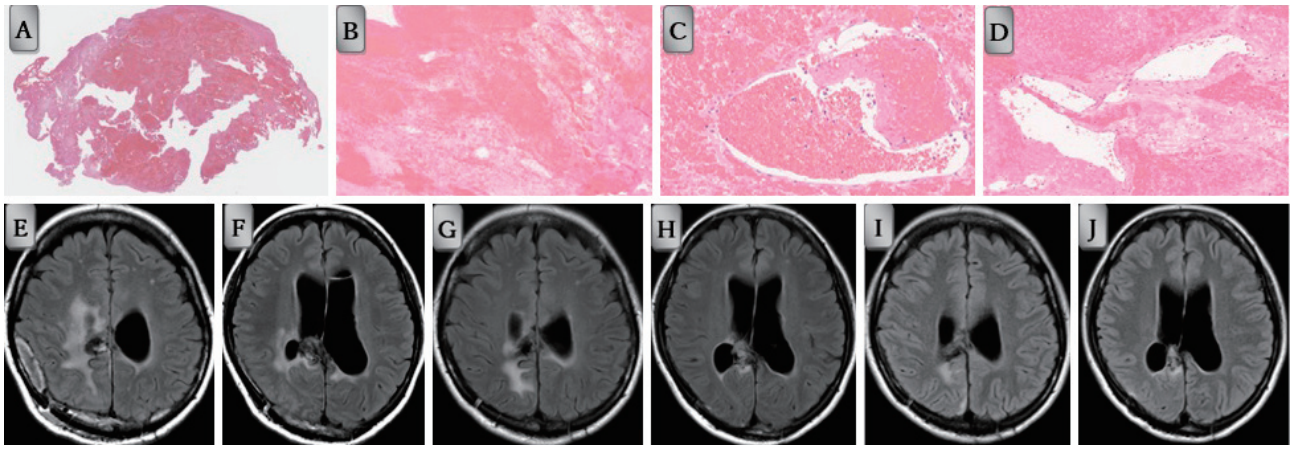

Fig. 4
(A–D): Pathology findings show the hematoma surrounded by a tough capsule and abnormal blood vessels with large and small dilatations within the hematoma. The lesions comprised endothelial cells and the surrounding thin layer of dense fibrous tissue and pericytes, leading to a pathological diagnosis of CEH.
(E, F): Axial FLAIR MRI obtained in 2 weeks after surgery.
(G, H): Axial FLAIR MRI obtained in 2 months after surgery.
(I, J): Axial FLAIR MRI obtained in 6 months after surgery. Edematous changes around the lesion improved markedly over time.
CEH: chronic expanding hematoma; FLAIR: fluid-attenuated inversion recovery; MRI: magnetic resonance image

layer of the capsule.[4] Recent reports suggest that vascular endothelial growth factor (VEGF) is likely involved in the formation of CEH.[1,4,5,8-11] VEGF is a specific regulator that promotes endothelial cell growth, proliferation, and differentiation.[4,5,8,12] Consequently, CEH is believed to expand through repeated angiogenesis and local hemorrhage.[1,4,5,8]

Surgical intervention is effective for treating CEH with neurological deficits.[1,5] Edematous changes around the CEH often improve rapidly after surgery, which helps to halt disease progression and significantly enhance the patient's neurological improvement.[1] Recurrence of residual lesion after partial removal has been reported, and gross total resection of the lesion is the curative treatment.[13] However, this is often difficult because of adhesions to the surrounding tissues.[14] In this case, symptoms and edema around the lesion were relieved after surgery. Because it is possible that residual lesions may recur in the future, careful follow-up will continue.

The high resolution provided by endoscopic surgery is particularly useful for removing deep-seated lesions.[5,15,16] CEH associated with AVM is located in deep structures, where GKS is often applied. However, endoscopic surgery, which offers a wide operative field with a panoramic view, is highly effective. Moreover, endoscopic surgery provides a visual field that is not achievable with microscopic surgery, enabling safer operations.[15,16] In this case, we could clearly visualize the dorsal side of the bilateral ICVs, which are difficult to visualize using the conventional microsurgical posterior interhemispheric approach. The safe operative field provided by endoscopic surgery likely contributed to a favorable postoperative course without complications. To our knowledge, this is the first report of endoscopic surgery for intracranial CEH. Endoscopic surgery becomes a treatment option with safe surgical corridor in the selected patients having deep-seated intracranial CEH.

Conclusion

We presented a case of delayed onset CEH after GKS for AVM, successfully treated with surgical intervention. The pathophysiology, optimal treatment, and timing of surgery for CEH remain unclear and require further research. In this case, endoscopic surgery was effective and safe for the removal of CEH. The bright and wide operative field provided by endoscopy has an advantage for CEH associated with deep-seated AVMs. In addition, endoscopic surgery offers an operative field beyond that achievable with traditional microscopic techniques, enabling safer and more effective surgical removal.

Acknowledgments

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Informed Consent

Informed consent was obtained from the patient.

Conflicts of Interest Disclosure

All authors have no conflict of interest. All authors are members of The Japan Neurosurgical Society and have registered online Self-reported Conflict of Interest Disclosure Statement Forms.

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