A Case of Recurrent Hemorrhages due to a Chronic Expanding Encapsulated Intracranial Hematoma

Akiko Marutani a Kiyoshi Nagata a Jun Deguchi a Yuji Nikaido a
Syuji Kazuki b

a Department of Neurosurgery, Nara City Hospital, and
b Department of Neurosurgery, Kazuki Clinic, Nara City, Japan

Key Words
Encapsulated intracranial hematoma · Fibrous capsule · Angiographically occult intracranial vascular malformation · Chronic encapsulated hematoma

Abstract
Few case reports of encapsulated intracranial hematoma (EIH) exist, and the mechanisms underlying the onset and enlargement of EIH remain unclear. Here, we report on a 39-year-old woman with an EIH that repeatedly hemorrhaged and swelled and was ultimately surgically removed. In June 2012, the patient visited her local doctor, complaining of headaches. A magnetic resonance imaging (MRI) scan identified a small hemorrhage of approximately 7 mm in her right basal ganglia, and a wait-and-see approach was adopted. Six months later, her headaches recurred. She was admitted to our department after MRI showed tumor lesions accompanying the intermittent hemorrhaging in the right basal ganglia. After admission, hemorrhaging was again observed, with symptoms progressing to left-sided hemiplegia and fluctuating consciousness; thus, a craniotomy was performed. No obvious abnormal blood vessels were observed on the preoperative cerebral angiography. We accessed the lesion using a transcortical approach via a right frontotemporal craniotomy and removed the subacute hematoma by extracting the encapsulated tumor as a single mass. Subsequent pathological examinations showed that the hematoma exhibited abnormal internal vascularization and was covered with a capsule formed from growing capillaries and accumulating collagen fibers, suggesting that it was an EIH. No lingering neurological symptoms were noted upon postoperative follow-up. This type of hematoma expands slowly and is asymptomatic, with reported cases consisting of patients that already have neurological deficits due
to progressive hematoma growth. Our report is one of a few to provide a clinical picture of the initial stages that occur prior to hematoma encapsulation.

Introduction

Chronic encapsulated intracranial hematoma (EIH) is a rare type of intracranial hematoma. Few case reports for this condition exist, and the pathology remains unclear. Here, we describe a patient with a recurrent microhemorrhage – which after 6 months transitioned to a chronic expanding EIH – on whom operative treatment was performed. Using magnetic resonance imaging (MRI) and histopathological investigations, we present useful information about the process of EIH formation and review the relevant literature.

Case Report

A 39-year-old female presented with progressive headaches over the previous 6 months. She did not have any medical history or family history. In June 2012, the patient had received a detailed examination for mild headaches by a local physician. MRI scans had revealed a small hemorrhage of approximately 7 mm located in the right basal nucleus; however, because her symptoms improved, the patient had been assigned for observation. The acute headaches located on the right side at the back of the head recurred 6 months later, and MRI scans revealed a hemorrhage associated with a mass lesion that was different from the hemorrhage originally found in the right basal nucleus. Therefore, the patient was referred to our hospital.

Upon admission, her blood was analyzed, and the blood chemistry examination and assessment for tumor markers revealed no apparent abnormalities. Neurological examinations at admission showed she was lucid with headaches, with no other neurological deficits. Her modified Rankin Scale score prior to the operation was 4.

Neuroradiological Findings

Magnetic Resonance Imaging

The physician who had originally treated the patient found a 5-mm hematoma through T1- and T2-weighted imaging, and found a 7-mm mass exhibiting a mixture of iso- and hyperintense areas in the right basal nucleus (fig. 1). After 6 months (the time of referral to our hospital), increases in lesion size and peripheral edema were noted. The central part of the lesion was irregular and had iso-to-hyperintense mixed signals. This combination of mixed signal intensities identified 2 hematomas: a comparatively new hematoma (deoxyhemoglobin) and a subacute-phase hematoma (methemoglobin) (fig. 1).

Upon admission to our hospital, a 40-mm mass was observed that extended from the right basal nucleus to the frontal lobe. T1 and T2 scans revealed caudal multilocular anachronous hemorrhages associated with cystic components in the mixed-signal region. Furthermore, T1- and T2-weighted imaging revealed chronic hematoma (hemosiderin) and edematous changes, while T2* images showed hematoma expansion with gadolinium-diethylenetriamine pentaacetic acid irregular enhancement effects.
Digital Subtraction Angiography
Frontal and lateral right coronary artery angiography of the arterial and venous phases did not show any malformed blood vessels, and no abnormal staining was noted. No apparent venation abnormalities were noted (fig. 2).

Computed Tomography
Upon admission, new and old hemorrhagic lesions were observed. One week after admission, hemorrhage and lesion expansion continued, putting strong pressure on the internal capsule.

Clinical Course
One week after hospitalization, imaging revealed recurrent hemorrhaging. Since her consciousness disturbances and left hemiplegia had progressed, we performed a craniotomy. Preoperative cerebral computed tomography (CT) scans revealed no apparent vascular abnormalities.

Intraoperative Findings
A right frontotemporal craniotomy was conducted using the transcortical approach, and a corticotomy was performed on the nearest brain surface site. While performing aspiration on the comparatively new hematoma located on the surface of the tumor, we further conducted a dissection of the deeper parts of the brain near the lesion. Upon reaching the tumor, the boundary of the tumor surface was well defined, and the hard capsule was dark red in color. Due to the presence of old and new hemorrhages, the neighboring tissues were yellowish brown in color. After aspirating the dark-red hematoma mass in the deep part of the tumor, the tumor region was carefully excised and removed en bloc.

Histopathological Findings
HE Staining
The inside of the hematoma was covered by a hard capsule, and dilated blood vessels of various sizes were densely accumulated inside. The capsule consisted of hyperplasia of the outer collagenous layer and capillary vessels. The structure was the same as the adventitia of a chronic subdural hematoma. In one area, an accumulation of blood vessels with weakened, dilated, and tortuous wall structures associated with hyperplasia of malformed vessels was noted. The encapsulated wall contained fibroblast hyperplasia and hemosiderin-laden macrophages. Furthermore, many malformed vessels characterized by thin walls (fig. 3) were observed.

Elastica van Gieson Staining
No elastic membrane was observed, and factor VIII, a marker of endothelial cells, was positive at Ab13 (fig. 3).

Collectively, the histopathological findings revealed that hematomas appearing at different times were encapsulated by collagen fibers and capillaries, and the surface of the neoplasm contained hyperplasia of malformed vessels; therefore, the patient was diagnosed with EIH. Upon pathological examination, no vascular malformations such as neoplastic lesions or cavernous angiomas were observed. Thus, it was thought that a venous hemangioma existed.
Postoperative Course

No neurological symptoms were noted in the patient postoperatively, and she was discharged 2 weeks later. Her modified Rankin Scale score improved from 4 (obtained before the operation) to 0 (acquired at discharge).

Discussion

EIH, a rare type of cerebral hemorrhage, was first reported in 1978 by Yashon and Konsik [1], and about 50 cases have been reported since then [2]. EIH is defined as an intracranial hematoma that sometimes grows progressively [3]. Compared with normal intracerebral hemorrhages, EIH has an early onset age and can be complicated by hypertension, and almost all of the lesions develop beneath the cerebral cortex [4]. Although many theories concerning the etiology [5, 6] and mechanisms [6–8] of the disease have been proposed, the details remain unclear; thus, only around 20% of cases are diagnosed preoperatively [3]. Most previous studies report on expanded encapsulated hematomas that were discovered after the manifestation of neurological symptoms, with only a few studies discussing the initial stage of the disease before encapsulated hematoma development. Although EIH is a progressive and benign disease, neuroradiological findings have not been useful in its diagnosis; thus, a definitive diagnosis without pathologically examining the surgically excised lesion is difficult.

Histological findings of EIH reported by Hirsh et al. [6] showed that vascular malformations due to fibroblasts are involved in capsule development. Patients with suspected vascular malformations tend to have angiographically occult intracranial vascular malformations (AOIVMs) upon histological examination [8–10]. After the development of CT, it has widely been reported that AOIVMs cause headaches, intracranial hemorrhaging, and refractory epilepsy [8]. The classification of AOIVMs into arteriovenous malformations, cavernous angiomas, capillary telangiectasias, and venous angiomas is widely accepted. Takeuchi et al. [11] reported that vascular endothelial growth factor might be involved in EIH development. Further, since both leakage from the new blood vessels and strong infiltration of lymphocytes are observed, it is likely that chronic inflammatory reactions are involved in the edema around the hematoma [11].

Steiger et al. [10] reported that AOIVM-related capsule formation is often observed in cases of cystic cavernous angioma, and while it has the same structure as EIH, it is characterized by prominent peripheral edema. This peripheral edema may mainly be caused by exposure of the blood vessels that possess endothelial gap junctions on the outer layer of the capsule. One theory attributes capsule formation to AOIVM-induced fibroblasts, while another theory states that capsules are due to granulation tissue rich in blood vessels, which develops as an overreaction to hematomas [12]. Furthermore, recurrent hemorrhaging from the capsule and the AOIVM itself may lead to hematoma expansion [10, 12]. With regard to operative treatment, AOIVMs cause recurrent hemorrhaging, which can have a fatal outcome. However, because the border of the neoplasm is well defined, after reaching the brain depth near the lesion, removal can be performed rather easily; therefore, intentional operative excision is recommended [13]. Lobato et al. [14] reported statistical data for cases where patients underwent uneventful successful operative treatment for AOIVM of the brain stem, cerebral ventricles, paraventricular region, and basal ganglia. In the present case, complete removal of the tumor in the basal ganglia was achieved without any postoperative neurological complications.
Based on the pathological findings in the present case, we believe that vascular malformations due to vascular components such as AOIVMs were the source of the initial hemorrhage. As a result, capsule formation developed and the hematoma continued to grow, causing multiple hemorrhages and EIH development [15]. The mechanism of growth is thought to involve repeated bleeding from the new blood vessels in the capsule and the vascular malformation itself.

**Conclusions**

Here, we reported on a patient with recurrent hemorrhages and expanding EIH. We speculate that vascular malformations such as AOIVMs – which were thought to be the vascular components – were the source of the initial hemorrhage. The information presented in our report may aid in the diagnosis of EIH, regardless of the early onset age, complication of hypertension, or development of lesions throughout the cerebral cortex. In the future, clinicians should acquire multiple MR images because of the progressive growth of the EIH.

**Statement of Ethics**

Written informed consent was obtained from the patient before surgery and treatment. Since this is a case report, ethics approval was not sought.

**Disclosure Statement**

There are no conflicts of interest.

**References**

1. Yashon D, Konsik EJ: Chronic intracerebral hematoma. Neurosurgery 1978;2:103–106.
2. Miyahara K, Fujitsu K, Yagihita S, Ichikawa T, Takemoto Y, Okada T, Niiro H, Shina T: Chronic encapsulated intracerebral hematoma associated with cavernous angioma – case report. Neurol Med Chir (Tokyo) 2011;51:52–55.
3. Nishiyama A, Toi H, Takai H, Hirai S, Yokosuka K, Matsushita N, Hirano K, Matsubara S, Nishimura H, Uno M: Chronic encapsulated intracerebral hematoma: three case reports and a literature review. Surg Neurol Int 2014;5:88.
4. Kasuya J, Hashimoto Y, Terasaki T, Miura M, Miyayama H, Uchino M: Chronic encapsulated intracerebral hematoma in thalamus with incongruous right homonymous hemianopia: a case report (in Japanese). Rinsho Shinkeigaku 2000;40:29–33.
5. Fiumura E, Gambacorta M, D’Angelo V, Ferrara M, Corona C: Chronic encapsulated intracerebral hematoma: pathogenic and diagnostic considerations. J Neurol Neurosurg Psychiatry 1989;52:1296–1299.
6. Hirsh LF, Spector HB, Bogdanoff BM: Chronic encapsulated intracerebral hematoma. Neurosurgery 1981;9:169–172.
7. Aoki N, Mizuguchi K: Chronic encapsulated intracerebellar hematoma in infancy: case report. Neurosurgery 1984;14:594–597.
8. Kumabe T, Kayama T, Sakurai Y, Ogasawara K, Niizuma H, Wada T, Namiki T: Encapsulated chronic intracerebral hematoma caused by venous angioma of the basal ganglia: a case report (in Japanese). No Shinkei Geka 1990;18:735–739.
9. Monma S, Ohno K, Hata H, Komatsu K, Ichimura K, Hirakawa K: Cavernous angioma with encapsulated intracerebral hematoma: report of two cases. Surg Neurol 1990;34:245–249.
10. Steiger HJ, Markwalder TM, Reulen HJ: Clinicopathological relations of cerebral cavernous angiomas: observations in eleven cases. Neurosurgery 1987;21:879–884.
Takeuchi S, Takasato Y, Masaoka H, Hayakawa T, Otani N, Yoshino Y, Yatsuhihe H, Sugawara T: Development of chronic encapsulated intracerebral hematoma after radiosurgery for a cerebral arteriovenous malformation. Acta Neurochir (Wien) 2009;151:1513–1515.

Pozzati E, Giuliani G, Gaist G, Piazza G, Vergoni G: Chronic expanding intracerebral hematoma. J Neurosurg 1986;65:611–614.

Yamasaki T, Handa H, Yamashita J, Paine JT, Tashiro Y, Uno A, Ishikawa M, Asato R: Intracranial and orbital cavernous angioma. A review of 30 cases. J Neurosurg 1986;64:197–208.

Lobato RD, Perez C, Rivas JJ, Cordobes F: Clinical, radiological, and pathological spectrum of angiographically occult intracranial vascular malformations. Analysis of 21 cases and review of the literature. J Neurosurg 1988;68:518–531.

Hideaki K, Toshihiro K, Teiji T, Kazuo M, Takashi Y: Multiple intracerebral hematoma representing a transitional form to chronic encapsulated hematoma: a case report. No Shinkei Geka 1997;6:555–559.

Fig. 1. MRI scans obtained at the onset of initial symptoms show the right basal nucleus iso-to-hyperintense area on T1- (left) and T2-weighted (right) images (top). MRI scans obtained 6 months after the onset of initial symptoms show the iso-to-hyperintense area surrounded by a hyperintense zone on T1- (left) and T2-weighted (right) images (bottom).
Fig. 2. Digital subtraction angiograms of the arterial (top) and the venous phase (bottom) are normal. Left: anteroposterior view. Right: lateral view.
Fig. 3. Photomicrograph of the capsule. There is hyperplasia of the vessel malformation. The inner layer of the tumor capsule shows old-to-fresh hematomas. There is a venous angioma surrounding the endothelial cells and smooth muscle layer. Top: HE; ×40. No elastic membrane is observed, and factor VIII, a marker of endothelial cells, is positive at Ab13. Bottom: Elastica van Gieson staining; ×40.