

A Case of Bilateral Cerebellar Chronic Encapsulated Intracerebral Hematoma with Rapidly Progressive Symptoms

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Abstract

Chronic encapsulated intracerebral hematoma (CEIH) is a rare cerebrovascular disease featuring progressively expanding intracranial hematoma. We treated a man in his 70s with bilateral cerebellar CEIH. He had presented at another hospital with dizziness, and imaging showed two independent hemorrhagic space-occupying lesions in the bilateral cerebellar hemispheres. The symptoms progressed relatively rapidly, and there were signs of impending cerebellar herniation; he was transferred to our institution, and emergency surgery was performed. The operative findings included a hematoma with partial capsulation. We diagnosed CEIH from preoperative magnetic resonance imaging and computed tomography findings, clinical course, and pathological findings. The postoperative course was satisfactory. We present this case of bilateral cerebellar CEIH, as an extensive search of the literature suggests that this has not been reported before. Although CEIH is a condition that is usually hard to diagnose preoperatively, good outcomes can be achieved with appropriate surgical treatment. It is therefore important to keep this clinical entity in mind and not miss the right timing to operate.

Keywords: chronic encapsulated intracerebral hematoma, bilateral cerebellar hemorrhage, intracerebral hemorrhage

Introduction

Chronic encapsulated intracerebral hematoma (CEIH) is a rare condition that presents as a progressive exacerbation of neurological symptoms over weeks to months from the onset with a slowly expanding hematoma.^{1,2)} Magnetic resonance imaging (MRI) often shows elliptical lesions containing hematomas with clear borders, surrounded by peripheral edema.³⁾ The capsule often shows as a low-intensity rim on a T2-weighted image.⁴⁾ Enhanced imaging sometimes shows ring enhancement.^{3,5,6)} CEIH with symptomatic progression was first described by Yashon et al. in 1978 and by Hirsh et al. in 1981.^{1,2)} Takahashi et al. reported a chronic multilocular encapsulated intracerebral hematoma several years later.⁷⁾ Since then, approximately 70 cases have been documented, mostly from Japan. In approximately 80% of cases, the CEIH is located in the subcortical region, but there are also reports of CEIH in other areas, including the cerebral deep region, cerebellum, and

cerebral ventricle.⁸⁾ There are no clear diagnostic criteria at present and CEIH is often difficult to diagnose especially preoperatively. Diagnosis of this clinical entity can be made only with preoperative images, intraoperative findings, and pathological findings. This time we performed a surgical evacuation of hematomas in a case with two simultaneous CEIH occurring bilaterally in the cerebellum. Multiple CEIH in the posterior fossa have not been reported so far. We report this case with a review of the literature.

Case Report

The patient was an Asian man in his 70s with a history of cholangiocarcinoma, early gastric cancer, emphysema, and hypertension and was taking oral low-dose aspirin after surgery for thoracic aortic aneurysm. Dizziness and lightheadedness appeared approximately 3 weeks before admission to our hospital, and the symptoms gradually

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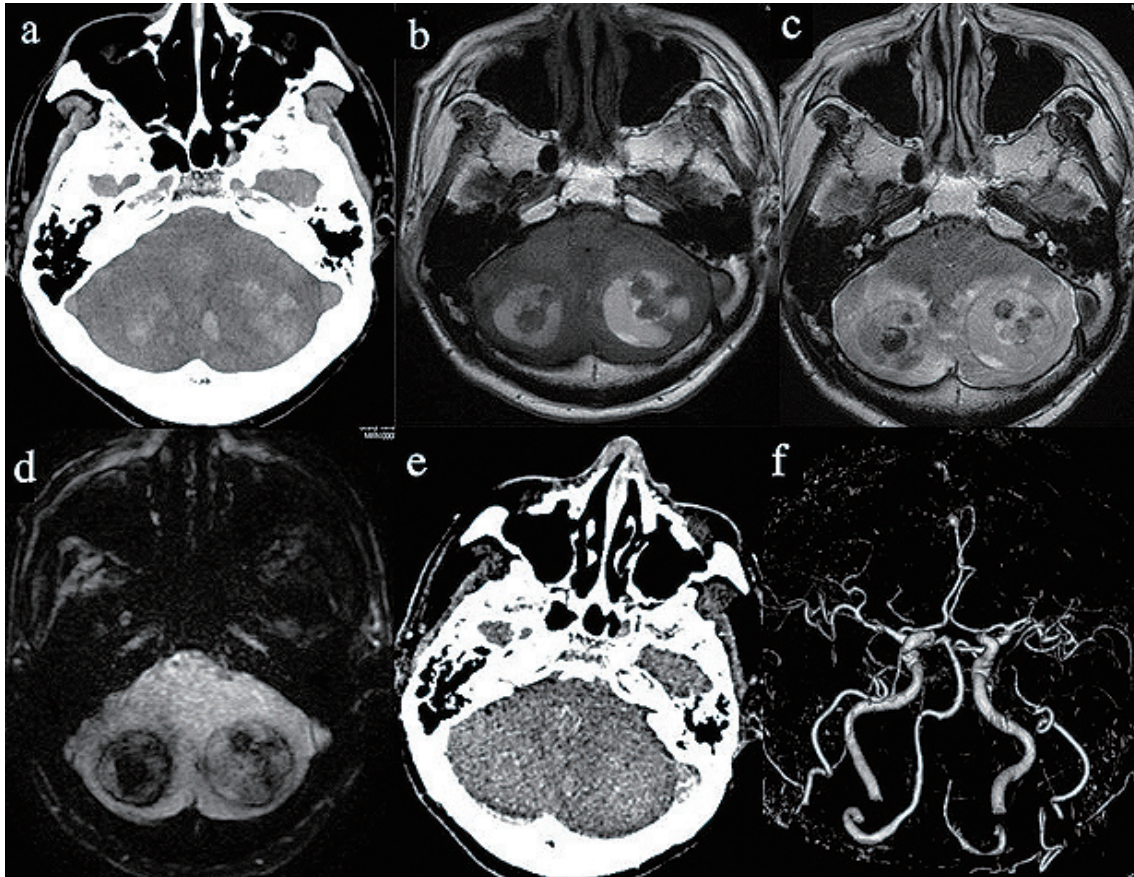


Fig. 1 Preoperative images.

Computed tomography¹⁾ showed high- and low-density areas with a maximum diameter of 31 and 34 mm, respectively, in the right and left cerebellum (a). T1-weighted (b), T2-weighted (c), and T2*-weighted MR (d) images showed the lesions with mixed signals. Perifocal edema was observed around the periphery, and T2WI and T2*WI showed a low-signal ring sign around the lesions. Enhanced CT showed no obvious abnormal blood vessels or tumor (e). 3DCTA showed cerebral aneurysm in the right distal ACA; however, no apparent vascular lesion was identified in the posterior fossa (f).

worsened; so, the patient visited a nearby doctor 1 week before admission to our hospital. Computed tomography¹⁾ showed areas with a maximum diameter of 31 and 34 mm, respectively, in the right and left cerebellum that exhibited high- and low-density patches and were more mottled than in a typical cerebellar hemorrhage (Fig. 1a). The patient was hospitalized with a diagnosis of intracranial hemorrhagic space-occupying lesions. The lesions showed a mix of high and low signals on T1 WI, T2 WI in MRI, and perifocal edema was observed around the periphery (Fig. 1b, c). Additionally, multiple cystic lesions with low signal in the interior as well as T2 WI and T2* WI low-signal ring signs around the lesions were observed (Fig. 1c, d). For a differential diagnosis, cavernous hemangioma was initially suspected because of the clear border of the lesions or metastatic brain tumor because of the history of early gastric cancer and bile duct tumor. Conservative therapy was initially chosen at a previous hospital because the patient had a clear level of consciousness and had only complained of dizziness and lightheadedness at the begin-

ning. The patient was planned to be transferred to our hospital where neurosurgical management including surgery was available. However, the symptoms gradually got worse a few days before the transfer. This was turned out when the patient was transferred as planned, and after transferring to our institution, the patient was determined to have disturbed consciousness (Glasgow Coma Scale E3V 3M5) on admission and his respiration was also unstable. Another CT scan was performed at our hospital, which revealed that the lesions had not significantly changed in size, but the density inside the lesions had changed from the previous CT and the edema around the lesions was exacerbated. The posterior fossa was tight, and the brainstem was compressed. Because of the need for urgent decompression surgery, only contrast-enhanced CT and 3D-CTA were performed to rule out vascular or neoplastic lesions, which did not show any of these findings (Fig. 1e, f). No cerebral angiography or contrast-enhanced MRI was performed. Laboratory results on admission showed a platelet count of $314 \times 10^3/\mu\text{L}$, APTT 28.8, and PT-INR 1.26.

The ARU on VerifyNow™ analysis was 536.

The surgery was performed under general anesthesia with the patient in a prone position. A longer-than-usual skin incision was made in the midline occipital region. An extensive bilateral suboccipital craniotomy was performed to expose the bone covering the posterior fossa. Inferiorly, the foramen magnum was also opened. The dura was opened with an X-shaped incision, initially just above the left cerebellum in the area where we expected to uncap.

After making corticotomy, a thin capsule was observed that could easily rupture when the lesion was reached, and a reddish-brown old hematoma was drained from the interior. When the hematoma was removed and decompressed, the dura was widely opened, and the lesion was extracted while confirming the border between the sur-

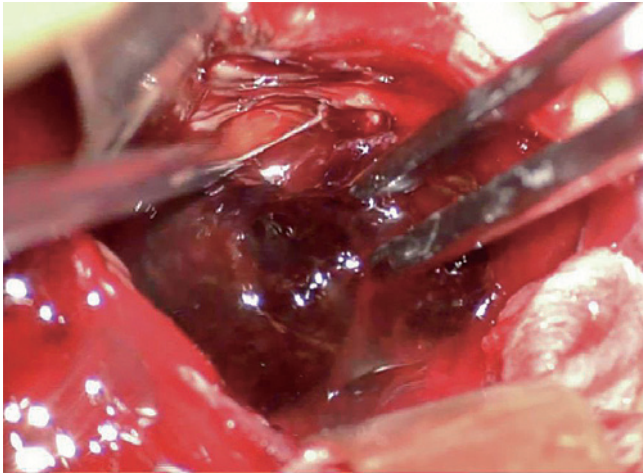


Fig. 2 Operative view.

The operative view of the lesion of the left side. It showed a thin capsule that could easily rupture, and a reddish-brown old hematoma was drained from the interior when the capsule was breached.

rounding normal brain tissue and the lesion. The lesion partly comprised nodular tissue with capsule and minute blood vessels entering from the periphery. These vessels were cauterized and cut, and the lesion was extracted (Fig. 2). After hemostasis of the left side was confirmed, the lesion on the right side was extracted in the same way. Similar findings were observed on the right side, and nodular tissue with capsule was removed from the surrounding brain. The volume of the posterior fossa was enlarged with an artificial dura mater, the bone flap was not put back in place, and surgery was completed with external decompression (Fig. 3a). Pathological findings showed multiple capillaries in hematoma, which can be taken as the possibility of secondary changes in response to hematoma (Fig. 4a-c). No tumor cells or vascular lesions such as cavernous hemangioma, arteriovenous malformation, or venous hemangioma were found. The lesions were diagnosed as CEIH on the basis of preoperative imaging, clinical course, and intraoperative findings. One week after surgery, the patient developed venous thrombosis of the leg and slight pulmonary thromboembolism. Heparin was started besides the low-dose aspirin that had been resumed after the aortic aneurysm operation. This led to subcutaneous hematoma in the wound area, and hematoma removal was required. The patient made good progress after the two operations, and the disturbance in consciousness improved. No recurrent cerebellar lesion was observed on CT and MRI taken 1 or 2 months later (Fig. 3b, c).

Discussion

CEIH often begins with progressive neurological symptoms due to a gradual increase in hematoma, unlike common intracerebral hematoma that presents with a sudden onset.^{1,4-6,8-12)} In the present case, after the symptoms of dizziness and lightheadedness had continued for approxi-

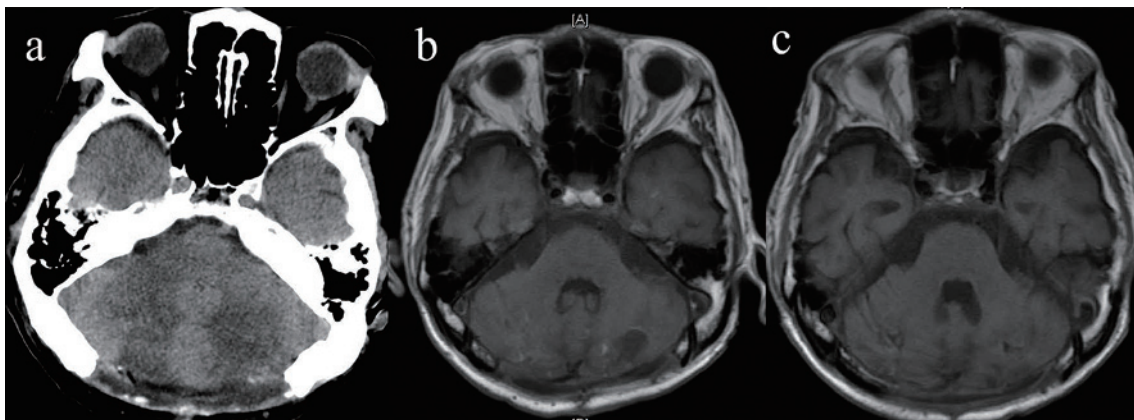


Fig. 3 Postoperative images taken after transferring to our hospital.

CT scan taken the next day of the operation showed that both lesions have been removed and external decompression was performed (a). T1-weighted MRI images taken 1 month (b) and 2 months (c) after the operation showed no recurrent cerebellar lesion.

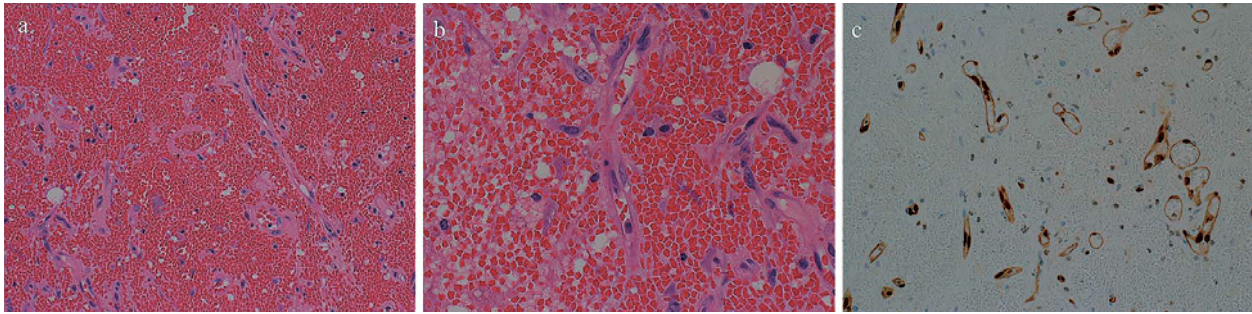


Fig. 4 Pathological findings.

Multiple capillaries in hematoma considered to be secondary changes in response to hematoma. No tumor cells or vascular lesions were found. Hematoxylin and eosin stain $\times 20$ (a), hematoxylin and eosin stain $\times 40$ (b), and capillaries were shown using ERG immunostaining (c).

Table 1 Reported cases of chronic encapsulated intracerebral hematoma in the cerebellum

Case no.	Author (year)	Age	Sex	Location	Side	Duration	Cause	Treatment	Outcome
1	Kawakami et al. (1978)	31	F	Cerebellum	lt	8 m	Unknown	Excision	Improvement
2	Aoki et al. (1984)	0	M	Cerebellum	mid	1 m	Unknown	Excision	Improvement
3	Masuzawa et al. (1985)	8	M	Cerebellum	lt	1 m	Unknown	Excision	Improvement
4	Aoki et al. (1986)	24	M	Cerebellum	rt	1 m	Unknown	Excision	Improvement
5	Matsumoto et al. (1988)	60	F	Cerebellum	lt	3 w	Carcinoma	Excision	Improvement
6	Takeuchi et al. (2008)	31	F	Cerebellum	lt	–	Unknown	Excision	–
7	Watanabe et al. (2014)	34	F	Cerebellum	lt	–	Arteriovenous malformations	Excision	Improvement
							SRS		
8	Present case	75	M	Cerebellum	bi	3 w	Unknown	Excision	Improvement

lt: left, mid: midline, bi: bilateral. SRS: stereotactic radiosurgery

mately 2 weeks, the symptoms gradually progressed over the course of 1 week. It took about 3 weeks from the onset for the hematoma and peripheral edema to worsen, and this led to impending cerebellar herniation. The clinical course is consistent with previous reports of CEIH.^{1,4,5,9,13-15} Preoperative MR image (T2 WI) showed a low signal for the envelope, whereas the center of the lesion showed mixed density with a clear border, which is also compatible with CEIH.³ We considered intratumoral hemorrhage of cavernous hemangioma or metastatic brain tumor as differential diagnoses, but the pathological findings ruled these conditions out. We therefore made a diagnosis of bilateral cerebellar CEIH.

The etiology of this disease remains unknown. Various causes of CEIH have been reported; vascular anomalies including cavernous hemangioma and radiotherapy are often the underlying cause, but not in this case.^{4,13,14,16-20} We consider that the pathogenesis of CEIH in this case is related to the cancer-bearing condition and antiplatelet therapy. The patient had a history of cholangiocarcinoma and early gastric cancer and was taking low-dose aspirin. Although there have been no reports of an association between

CEIH and cancer-bearing conditions, several reports suggested the relationship between intracranial hemorrhage (ICH) and cancer-bearing patients. Cerebrovascular events are common in patients with cancer, with ICH accounting for approximately half of these events.²¹⁻²³ In patients with cancer, ICH is caused by a complex mechanism in malignancy, including coagulopathy, neovascularization, or over-expression of factors such as vascular endothelial growth factor and matrix metalloproteinases.^{21,23-27} Among them, coagulopathy is a major factor in the development of ICH and can occur even in the early stages of cancer.^{21,28} It has also been reported that ICH due to coagulation disorders does²⁶ not necessarily indicate abnormal coagulation by blood sampling.^{26,28-31} In addition, in this case, the result of ARU measured using the VerifyNowTM system was <550 , suggesting that platelet aggregation was suppressed.

Additionally, an association between perihematomal edema and cancer has been reported. Gusdon et al. reported that patients with active cancer had larger perihematomal edema. They hypothesize that the immune system under cancer-bearing conditions results in a more robust inflammatory response to ICH, leading to more peri-

hematomal edema.³²⁾

Only seven cases of CEIH in the cerebellum have been reported thus far (Table 1).^{4,9,12,14,16,17,20)} The duration from initial onset to surgery is approximately 1 month, including the present case; this is a shorter period of time than in cases in other locations. This may originally be due to the lesion occurring in the posterior fossa with limited space. An extensive search of the literature suggests this is the first report of CEIH involvement occurring simultaneously in the bilateral cerebellum. We think that the present case with bilateral involvement resulted in a relatively faster progression of symptoms compared with cases involving one lesion. Since good outcomes have been reported in most postoperative cases, it is important to not miss the right timing to operate. CEIH should be considered as a differential diagnosis for intracranial space occupational lesions. In progressive conditions of CEIH, the gradual increase of hematoma with peripheral edema can be life-threatening, especially with the lesion in the posterior fossa. Especially, patients at risk for coagulation abnormalities should be closely monitored.

Conflicts of Interest Disclosure

All authors declare that they have no competing interests. The authors obtained written informed consent for this case report from the patient.

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