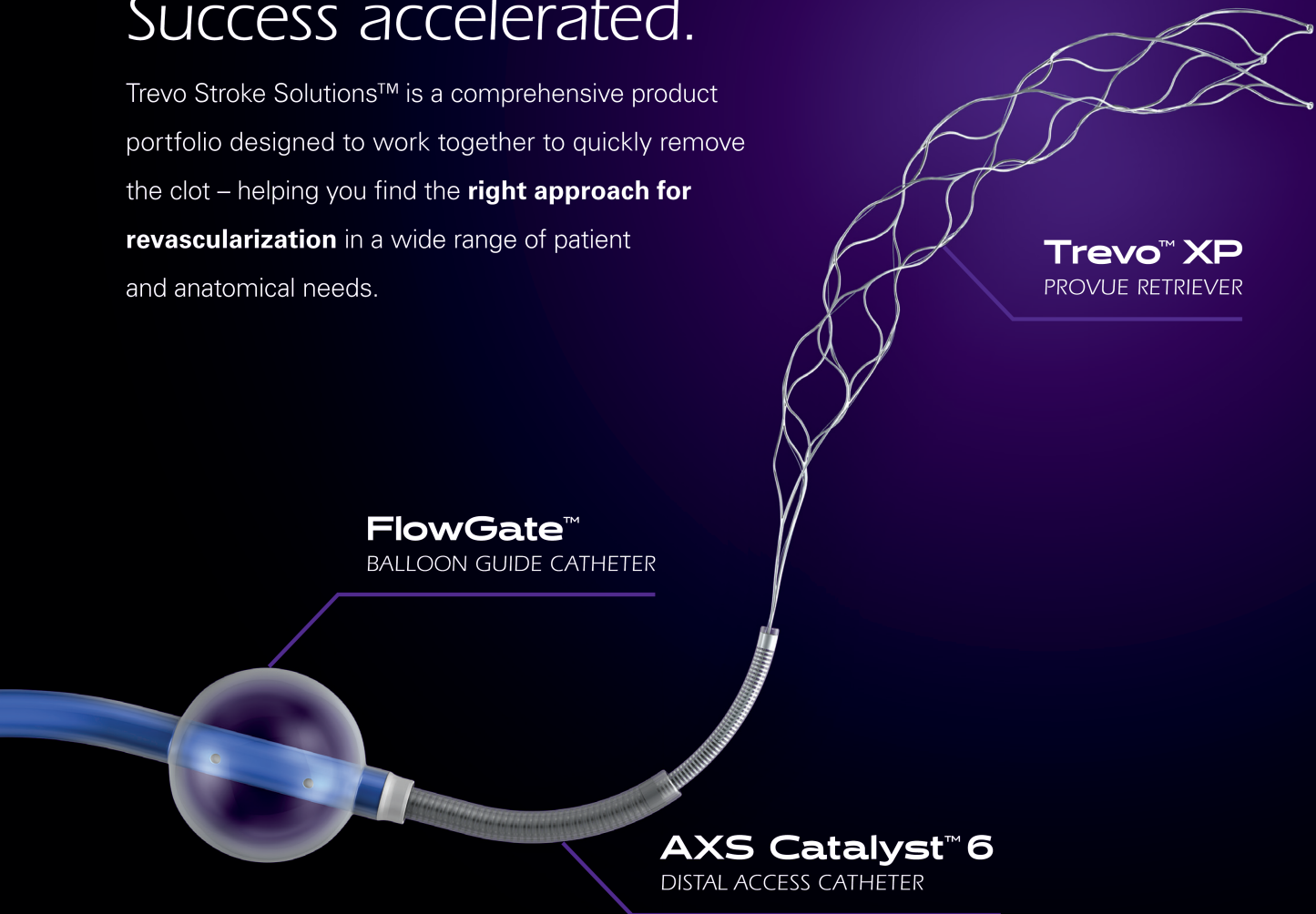


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Subarachnoid and intracerebral hemorrhage from intracranial hemangiopericytoma: An uncommon cause of intracranial hemorrhage

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Abstract

Background: Hemorrhage from an intracranial tumor is well known but uncommon. In cases of subarachnoid hemorrhage, aneurysm rupture is a main cause.

Case description: A 64-year-old woman presented with sudden-onset motor aphasia. Computed tomography revealed subarachnoid hemorrhage, and intracerebral hemorrhage in the left temporal lobe. From the findings of pre-enhancement computed tomography, hemorrhage from a left middle cerebral artery aneurysm was initially suspected. Further radiological examinations demonstrated an enhanced mass with dural attachment in the left temporal region, but no vascular abnormality. Emergency craniotomy was performed, and the tumor and intracerebral hematoma were removed. The hemorrhage from fragile tumor vessels may destroy the tumor tissue and spread into the temporal lobe and subarachnoid space. Pathological examination of the tumor yielded findings consistent with hemangiopericytoma. Post-operatively, the patient was treated to prevent vasospasm.

Conclusion: Although an intracranial tumor including hemangiopericytoma uncommonly causes subarachnoid hemorrhage, it should be taken into consideration as a source of intracranial hemorrhage.

Keywords

Subarachnoid hemorrhage, intracerebral hemorrhage, hemangiopericytoma, brain tumor

Introduction

Subarachnoid hemorrhage (SAH) and intracerebral hemorrhage (ICH) from brain tumor are known phenomena, but uncommon.¹ SAH commonly occurs due to rupture of an intracranial aneurysm. In addition, hypertension is a main cause of ICH. Therefore, for cases of SAH or ICH, we tend to focus on examinations for vascular anomalies.

Hemangiopericytoma is a rare tumor derived from pericytes located around capillaries.^{2,3} It shows radiological features similar to meningioma. Commonly, an intracranial tumor including hemangiopericytoma causes symptoms due to a mass effect. Intracranial hemangiopericytoma presenting with intracranial hemorrhage is very rare. To our knowledge, 12 cases of intracranial hemorrhage from hemangiopericytoma have been reported.^{2,4–8} Among them, only one case developed SAH.⁴ Others included nine cases of ICH and two of subdural hematoma.⁴ The present case is the second reported case of hemangiopericytoma presenting with SAH. In this report, we describe a patient with hemangiopericytoma which caused SAH and ICH. For this case, sufficient attention was not initially paid

to the faintly high-density lesion observed on the pre-enhancement computed tomography (CT). We discuss the radiological findings, clinical course and management of this uncommon condition.

Case presentation

A 64-year-old woman was admitted with sudden-onset motor aphasia. CT revealed SAH, and ICH in the left temporal lobe (Figure 1(a)). SAH was found in the basal cistern and spread to the right Sylvian fissure. SAH and ICH due to ruptured cerebral aneurysm on the left middle cerebral artery (MCA) was initially suspected. To detect vascular anomalies, three-dimensional CT angiography (3D-CTA) was performed. 3D-CTA

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failed to demonstrate any vascular anomalies. Raw 3D-CTA data demonstrated an enhanced extra-axial mass adjacent to the hematoma (Figure 1(b)). Angiography showed no vascular lesions including cerebral aneurysms other than an anomalous left duplicated MCA (Figure 1(c)).

Although faint tumor staining was observed, a feeding artery or early venous filling was not seen on angiography. The pre-operative diagnosis was meningioma presenting with SAH and ICH. Emergency craniotomy was performed, and the tumor and ICH were removed. The tumor was attached to the dural membrane. The tumor tissue was soft and suckable by aspirator. The tumor was well demarcated and detachment of the tumor from brain tissue was not difficult. No thick feeding artery was identified. A small amount of intra-tumoral hemorrhage was observed. Pathological examination revealed a highly cellular tumor with numerous mitosis and nuclear polymorphism (Figure 2(a)) accompanied by dilated staghorn-type vessels (Figure 2(b, c)). The MIB-1 index was 9.7%. Immunostaining for CD34 was positive in tumor and

endothelial cells (Figure 2(b)). The tumor cells were positive for vimentin (Figure 2(c)), but negative for EMA (Figure 2(d)). These pathological findings were consistent with the diagnosis of hemangiopericytoma. Post-operatively, the patient was treated with fasudil hydrochloride hydrate and sodium ozagrel, and a hypertensive and hypervolemic state was maintained to prevent vasospasm, as is done for common SAH cases. Her post-operative course was uneventful. She was transferred to another hospital for rehabilitation with mild cognitive impairment on the 38th day.

Discussion

Rupture of an intracranial aneurysm is commonly the main cause of SAH. Therefore, an aneurysm or vascular anomaly is initially sought by radiological examinations in cases of SAH. Intracranial hemorrhage from a brain tumor is well known. The incidence of hemorrhage from intracranial tumors is around 5%.⁵ Intracranial hemangiopericytoma is a rare tumor with malignant features which represents about 2% of all

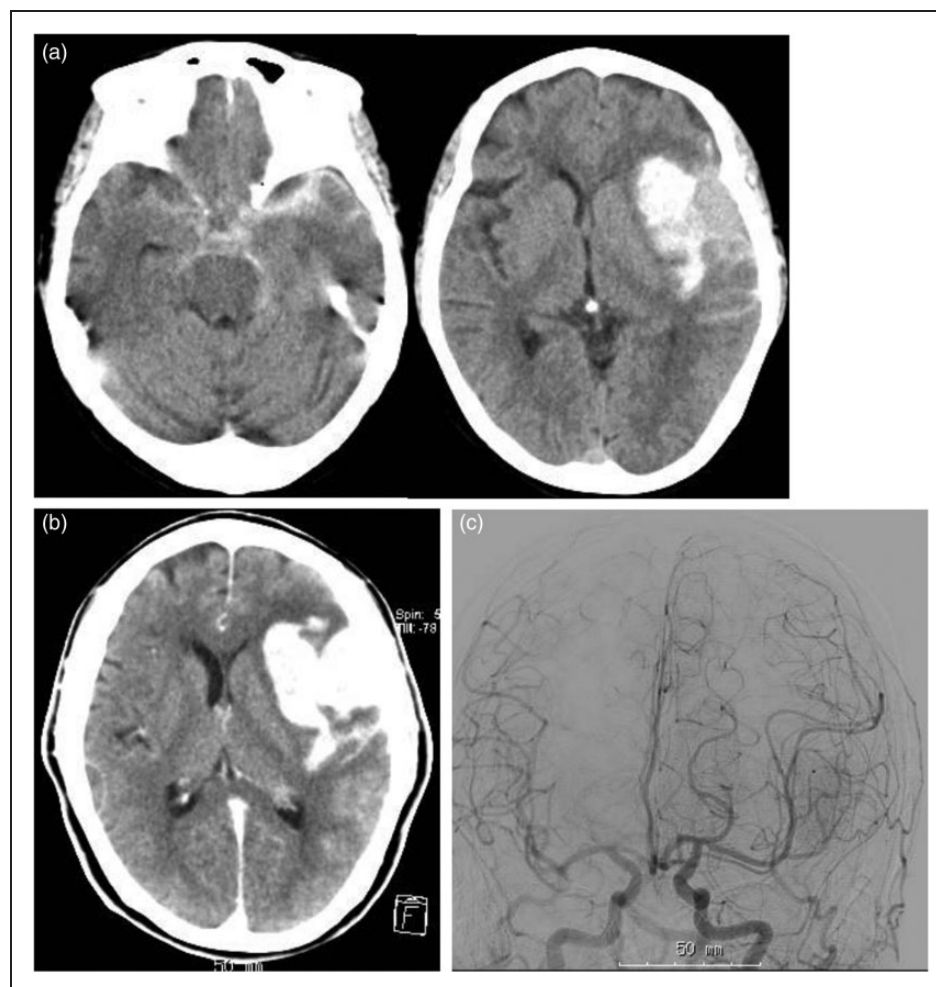


Figure 1. (a) CT on admission showing SAH, and ICH in the left temporal lobe. (b) Contrast enhanced-CT showing enhancement of an extra-axial mass in the left temporal region. (c) Angiography obtained under compression of the right carotid artery showing faint tumor staining, but not vascular lesions which can cause SAH or ICH.

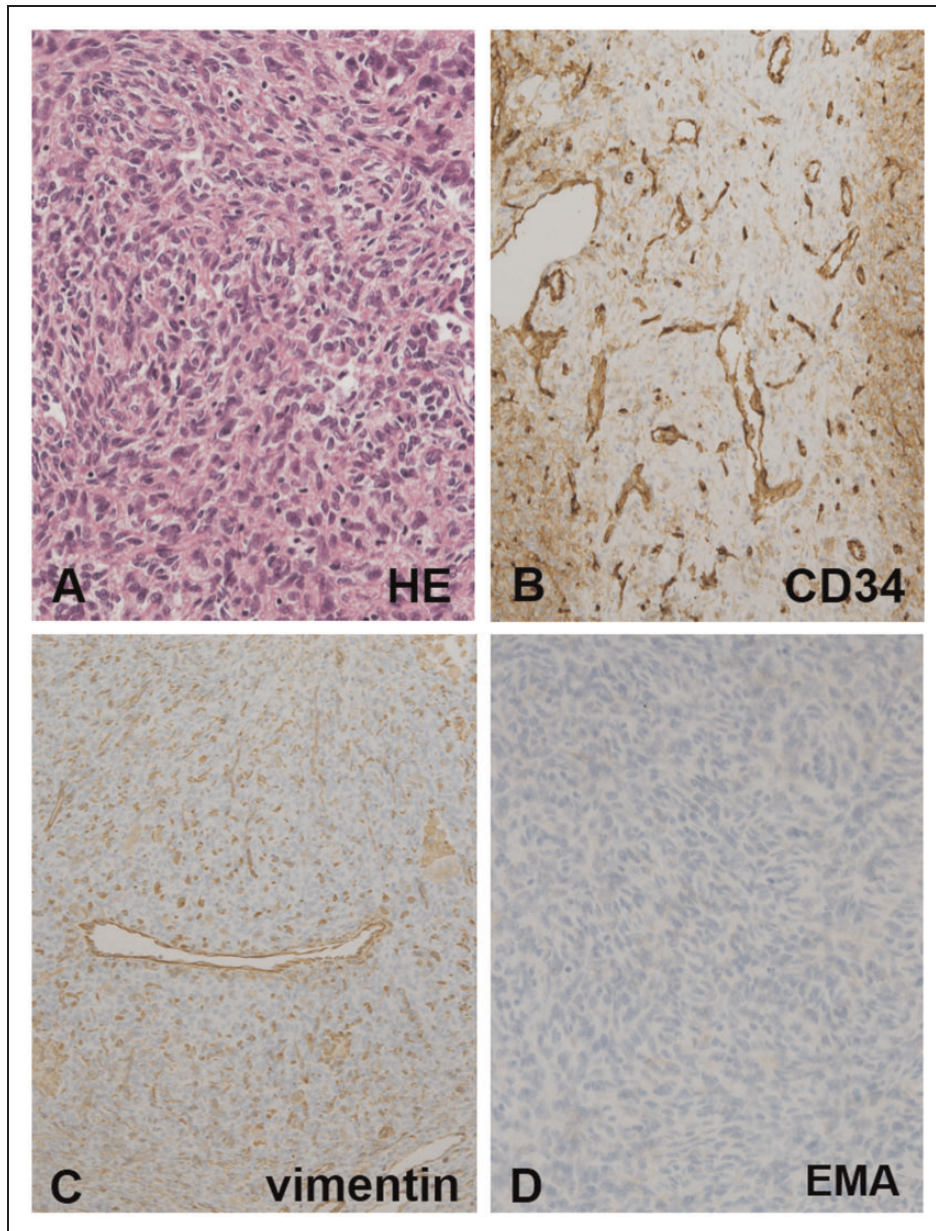


Figure 2. (a) A microphotograph of the specimen demonstrating the tumor cells with numerous mitoses. H & E, original magnification $\times 200$. The tumor cells were positive for CD34 (b) and vimentin (c), but not EMA (d). Original magnification $\times 100$. Panels B and C showing the compressed thin-walled blood vessels.

meningeal tumors and 1% of intracranial tumors.⁵ Up until 2014, a total of 12 cases of hemangiopericytoma presenting with intracranial hemorrhage had been reported.⁵ Among them, only one patient had SAH.⁴ Here, we report an additional case of this rare tumor presenting with SAH and ICH in which a hemorrhage due to a ruptured aneurysm was initially suspected.

Concerning radiological findings, CT on admission showed SAH spreading to the contralateral Sylvian fissure in addition to ICH. The tumor was demonstrated as a faintly high-density mass adjacent to the hematoma. However, sufficient attention was not paid to the lesion. Initially, rupture of the left MCA aneurysm was suspected based on the findings of pre-enhancement CT. Existence of a vascular lesion which could

cause SAH and ICH was investigated by 3D-CTA and angiography. As a result, raw 3D-CTA data demonstrated an enhanced mass attaching to the dural membrane in the left temporal region. After recognition of the tumor, the lesion was thought to be a meningioma. Pathological findings indicated that the tumor was consistent with hemangiopericytoma. There are some reported cases of meningiomas presenting with SAH.¹ Although SAH or ICH from an intracranial tumor is not common, we should be aware that a meningeal tumor might cause intracranial hemorrhage.

As for hemangiopericytoma, histological examinations have revealed that 23.4% of intracranial hemangiopericytoma showed intra-tumoral

hemorrhage microscopically.⁹ In hemangiopericytoma, blood vessels with erosion, distortion and distension caused by tumor growth, changes in the vessel wall structure, and occlusion and necrosis due to endothelial proliferation are considered to be possible sources of hemorrhage.⁵ In our case, intra-tumoral hemorrhage was observed during the operation, and the tumor was located in the temporal region with dural attachment and faced to the Sylvian fissure. Therefore, it is speculated that the hemorrhage from fragile tumor vessels may destroy the soft tumor tissue, and spread to the temporal lobe and subarachnoid space. Depending on the direction of bleeding, the location of the hemorrhage may vary.

Concerning symptoms and management, intracranial tumors with hemorrhage can lead to sudden-onset symptoms such as cerebral stroke. In our case, the patient developed motor aphasia. Based on the sudden-onset symptom and radiological findings, stroke was strongly suspected and examinations to detect vascular anomalies were done first. The hemorrhage was finally revealed to be from the tumor. For such cases of hemorrhage from an intracranial tumor, an emergency operation and post-operative treatment to prevent vasospasm are necessary, as for common SAH cases.

Conclusion

Although intracranial tumors with hemorrhagic onset remain rare, we should be aware of the existence of SAH and ICH from an intracranial extra-axial tumor such as hemangiopericytoma.

Contributions of authors

Shunsuke Seki: drafting of manuscript, critical revision, treatment of patient; Tomoya Kamide: critical revision, treatment of patient; Akira Tamase: critical revision, treatment of patient; Kentaro Mori: critical revision; Kunio

Yanagimoto: pathological examination; Motohiro Nomura: study conception and design, drafting of manuscript, critical revision, treatment of patient.

Conflict of interest

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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