

Intracerebral Capillary Hemangioma: A Case Report¹뇌 내 모세혈관성 혈관종의 증례 보고¹Inyoung Youn, MD¹, Jae Kyun Kim, MD¹, Jun Soo Byun, MD¹, Eon Sub Park, MD²Departments of ¹Radiology, ²Pathology, Chung-Ang University Medical Center, Chung-Ang University College of Medicine, Seoul, Korea

Intracerebral capillary hemangiomas are very rare benign vascular tumors that mostly occur during infancy. We described a 69-year-old man with generalized tonic-clonic seizures who was diagnosed with an intracranial mass. Multidetector computed tomography, magnetic resonance imaging and digital subtraction angiography studies were performed for evaluation of brain, and there was a well-enhancing mass found in the right temporal lobe without a definite feeding vessel. The patient underwent surgery and the pathologic examination demonstrated marked proliferation of small vessels with a lobular pattern in the brain parenchyma, which was confirmed to be capillary hemangioma.

Index termsHemangioma, Capillary
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INTRODUCTION

Capillary hemangiomas (CH) are benign vascular tumors or tumor-like lesions that usually occur in the skin, subcutaneous tissues, and mucous membranes of the oral cavities and lips. They may also occur in internal visceral organs such as the liver, spleen, and kidneys. The lesion most commonly occurs during infancy and usually regresses spontaneously with the passing of time; capillary hemangiomas occurring in the brain parenchyma are extremely rare. To our best knowledge, only 14 cases of CH arising in the brain have been reported with proven histology in the literature (1). We herein report on a rare case of cerebral capillary hemangioma in a 69-year-old man with generalized tonic-clonic seizure and review of the imaging findings of computed tomography (CT), magnetic resonance imaging (MRI) and digital subtraction angiography (DSA) studies.

CASE REPORT

A 69-year-old man initially presented with seizure-like movement of general tonic-clonic type and deviation of his eyeballs to the right side for 1 minute; he was admitted to our institute for further evaluation. He had experienced several instances of febrile seizures five or six years ago and had one more event about one year ago. The neurologic, physical and laboratory examinations had revealed no significant abnormality when he arrived at the hospital.

Multidetector CT of the brain and CT angiography was performed. On an unenhanced scan, the lesion was not clearly detected. After injection of contrast media, an approximately 12 mm, highly vascular mass lesion, was noted in the right temporal lobe. The lesion showed irregular focal dense enhancement and margin and was microlobulated. There was no related hemorrhage or calcification (Fig. 1A, B).

On a subsequently obtained MRI, the lesion showed intermediate signal intensity on T1-weighted images (T1WI) and hyperintensity on T2-weighted image (T2WI). In addition, the lesion was markedly hyperintense on T2-fluid attenuated

inversion recovery images and there was no signal void on susceptibility-weighted imaging (SWI). After administration of gadolinium-dimeglumine, the lesion showed intense spongiform enhancement and as a result, the diffusivity was in-

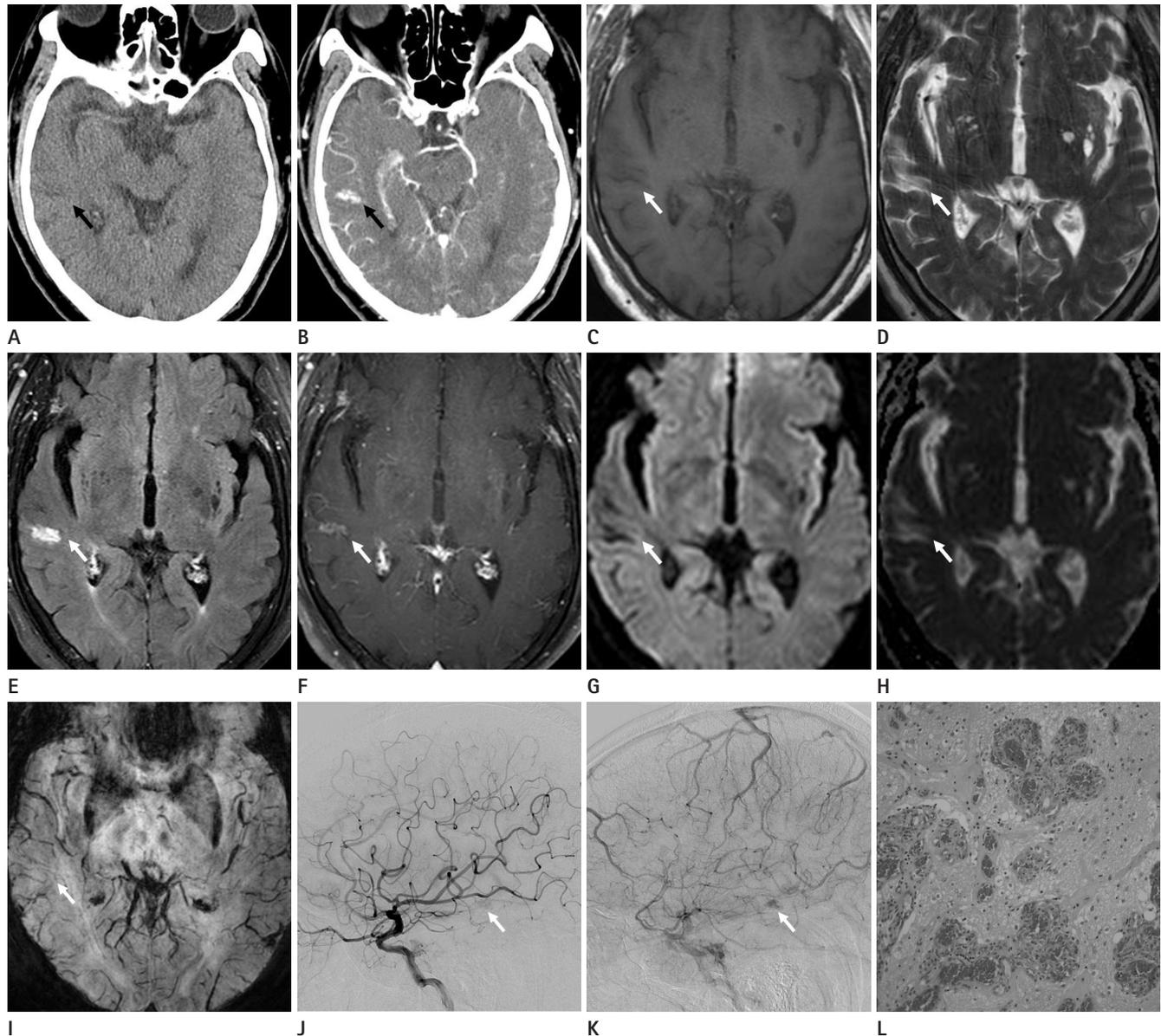


Fig. 1. A 69-year-old man with an intracerebral capillary hemangioma in right temporal lobe. **A, B.** Precontrast CT scan (**A**) showing the inconspicuous nature of the lesion (black arrow). Postcontrast CT scan (**B**) demonstrating strong nodular enhancement of the lesion (black arrow). **C-I.** The axial T1- (**C**) and T2- (**D**) weighted MR images subtly showing the mass lesion. The T2-FLAIR images (**E**) showing a more conspicuous appearance of the lesion (white arrow). Contrast enhanced images (**F**) demonstrating strongly spongiform enhancement of the lesion (white arrow). The DWI (**G**) and ADC map (**H**) of MRI demonstrating increased mean diffusivity and SWI (**I**) showing no signal void. **J, K.** Late arterial phase of DSA study (**J**) showing the beginning of contrast filling in the lesion (white arrow). Delayed phase (**K**) showing persistent staining similar in appearance to a puddle. These findings suggest a hypervascular tumor rather than a vascular malformation. **L.** The hematoxylin and eosin stained section ($\times 200$) reveals that lobulated but unencapsulated aggregates show closely packed, thin-walled capillaries, which are blood-filled and lined by a flattened endothelium, and separated by connective tissue stroma. **Note.**—ADC = apparent diffusion coefficients, DSA = digital subtraction angiography, DWI = diffusion weighted image, FLAIR = fluid attenuated inversion recovery, SWI = susceptibility-weighted imaging

creased on diffusion-weighted imaging (Fig. 1C-I). On a DSA study, the lesion showed delayed persistent staining without a feeding artery or draining vein (Fig. 1J, K). The differential diagnoses for a well-enhancing mass lesion without peritumoral edema were as followed: ganglioglioma, pilocytic astrocytoma, lymphoma, hemangioblastoma or hemangioma. Hemangioma or other vascular tumors should have been considered as part of the differential diagnoses because it showed intense enhancement suspected on CT and MRI scans and delayed persistent staining on DSA.

The patient underwent surgery because a glioma could not be excluded and a dark-reddish vascular mass was found in right temporal lobe without a feeding vessel on operation. The pathologic examination revealed proliferation of small vessels with a lobular pattern, which was lined by endothelial cells. The final histologic diagnosis was a capillary hemangioma (Fig. 1L). After discharge, the patient experienced no significant postoperative complications nor the recurrent seizure-like movement or other neurologic deficits for one year of follow-up.

DISCUSSION

CH are benign vascular tumors commonly occurring in full-term neonates, with an estimated frequency of 10 to 12% within the first year of life (2). The majority of CH arise in the skin and most of all show spontaneous regression without treatment. General imaging findings of CHs are different according to their stage of their natural history, that is, the proliferative phase or the involuting phase. In the proliferative phase, CH was a homogeneous soft tissue mass which showed a well-circumscribed margin, lobular contour, and intense and persistent enhancement on CT and MRI. The lesion also showed intermediate or decreased signal intensity on T1WI and moderately increased signal intensity with flow voids on T2WI. Heterogeneous signal-intensity, was occasionally seen, and may reflect thrombus or prior hemorrhage. Peritumoral edematous changes have not been noted. In the involuting phase, CH showed a decreased degree of enhancement and high-flow vascularity. Finally, CH was replaced by fibro-fatty tissue (3, 4). The enlarged arterial feeder and intense contrast staining was noted with the contrast pooling of DSA, and was probably due to the early draining vein or unambiguous arte-

rio venous shunting.

The hemangiomas are classified as either capillary hemangiomas, strawberry hemangiomas, or cavernous hemangiomas by the diameter of involving vessels (5). CHs are made up of capillary-sized vessels, while cavernous hemangiomas are composed of large dilated blood-filled vessels which often show thrombosis, perivascular hemosiderin deposition and calcifications (6). In the central nervous system, the cavernous hemangiomas more commonly occurred than CHs, especially in the cerebral hemispheres, and are usually found in older children and adults (7). Hemangiomas occurring in the cavernous sinuses have been reported and cavernous or mixed cavernous-capillary hemangiomas are more common than pure CHs (8). The cavernous hemangiomas of the cavernous sinuses are well-defined mass lesions that are strongly enhancing on CT scans. On MRI, cavernous hemangiomas are hypointense on T1WI and are very hyperintense on T2WI. After gadolinium administration, cavernous hemangiomas show strong and homogenous enhancement (7).

Intracerebral CH occurs extremely rarely and its pathogenesis, behavior, and natural history are not well understood (1, 2). Abe et al. (6) reported that central nervous system CH is similar to lobular CH of the skin, based on its histological and immunohistochemical features. Lobular CH is evenly distributed among all ages and developed rapidly, reaching its maximum size within weeks or months. As the apoptotic index of lobular CH is lower than that of infantile hemangioma, spontaneous involution of a central nervous system CH is not likely (6).

To our knowledge, there have been only a few reports of intracerebral CH confirmed with pathologic examinations. Intracerebral and intraspinal CH have been shown to exhibit the same cross-sectional imaging characteristics as cutaneous hemangiomas (4). Uyama et al. (1) reported that intracerebral CH occur in younger patients than in cases of intraspinal CH. The imaging findings of CH were demonstrated in a few reports as slightly high-attenuated areas enhanced by contrast medium on CT scan. While on MRI, the lesion showed iso- or hyperintensity areas on T1WI with marked enhancement and hyperintensity areas on T2WI. On the angiographic study, the hypervascular lesion was fed by pial, meningeal or branches of the middle cerebral artery, which were similar to those of meningioma (2, 6). The reported cases of intracerebral CHs

were accompanied by neurologic symptoms such as seizures, headaches, and nausea. The symptoms improved and there was no visible evidence of tumor recurrence after resection of tumors in most cases.

In our study, the lesion showed hypervascular mass in the right temporal lobe on CT scan. On MRI, the lesion showed intermediate signal intensity on T1WI with marked enhancement and hyperintensity on T2WI. Differential diagnoses for intracerebral CH include ganglioglioma, pilocytic astrocytoma, lymphoma, and hemangioblastoma. A ganglioglioma can be shown to be similar in appearance to a CH, but highly vascular enhancement is unusual. A pilocytic astrocytoma usually occurs in children. A primary central nervous system lymphoma typically shows decreased T2 signal intensity and apparent diffusion coefficients value as a result of its high cellularity. A flow void can be a clue of the hemangioblastoma. Intraparenchymal meningioma and intraparenchymal schwannoma can be present like a CH, but it is hard to include them in a differential diagnosis list because of their rarity.

There was no signal void in the lesion on SWI, which shows exquisite sensitivity to the venous vasculature, blood products, and vascular malformations (9). We assume that little amount of blood products within closely packed small vessels of CH, unlike large vascular channel filled with rich blood products in cavernous hemangiomas, caused insignificant results on SWI.

Extracutaneous CH involving multiple organs can exist anywhere in the body. Although most CH regresses spontaneously, some can have serious complications due to the capacity of unexpected tumor growth and hemorrhage (1). In particular, for cases of intracerebral CH, the complications can be fatal. Therefore, it is necessary to treat CH for prevention of a threatening course. In some cases, surgery may be also required.

We report here a case of intracerebral CH in a 69-year-old man presenting with seizure-like movement. The diagnosis of intracerebral CH was difficult to make because its characteristics are largely unknown due to its rarity. However, on the basis of our presenting case, a CH may be suggested in the case of hypervascular mass with strong spongiform enhancement on

CT or MRI and persistent staining on DSA. Well-ordered studies are necessary to characterize diagnostic imaging findings of intracerebral CH.

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뇌 내 모세혈관성 혈관종의 증례 보고¹

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뇌 내 모세혈관성 혈관종은 매우 드문 양성 혈관종양으로 대개 영아기에 발생한다. 우리는 뇌종양이 진단된 전신 긴장-간대성 발작을 보인 69세 남자 환자의 증례를 보고한다. 뇌의 평가를 위해 컴퓨터단층촬영, 자기공명영상, 그리고 디지털감산혈관조영술 검사가 시행되었고, 우측 측두엽에 영양혈관이 없는, 조영증강이 잘 되는 종양이 발견되었다. 수술적 치료가 시행되었으며, 병리적 소견상 뇌조직 내에 소엽상 소혈관의 과다 증식이 관찰되었고, 모세혈관성 혈관종으로 확진되었다.

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