Cavernous Angioma Associated with Venous Angioma
—Two Case Reports—

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Abstract
A 17-year-old male and a 28-year-old female presented with cavernous angioma and venous angioma colocated in the same region, and manifesting as convulsions. The cavernous angiomas were extirpated without damage to the venous angioma. No postoperative neurological deficits were observed. The patients have been free of convulsions without anticonvulsant medication for 2 years. Early extirpation of cavernous angioma with presentation of venous angioma is recommended, preferably before bleeding occurs.

Key words: cavernous angioma, venous angioma, vascular malformation, surgery

Introduction
Cerebral vascular malformations can be histologically classified into four categories: arteriovenous malformation, cavernous angioma, venous angioma, and capillary telangiectasia. This classification is widely adopted in clinical practice. Cavernous angioma, which sometimes causes convulsion or bleeding, is commonly considered by many to be an indication for operative extirpation. Angiographically occult vascular malformation requiring surgery is frequently associated with cavernous angioma. Venous angioma, which does not cause symptoms, may be of little clinical significance, although controversy remains. In addition, venous angioma is involved in normal medullary venous drainage, and extirpation results in brain swelling, so surgical extirpation is inadvisable. However, vascular malformation accompanied by venous angioma may cause symptoms such as bleeding and convulsion.

We describe two cases of cavernous angioma and venous angioma located in the same region, in which only the cavernous angioma was safely extirpated.

Case Reports
Case 1: A 17-year-old right-handed male was admitted to our hospital suffering from convulsions. No abnormal neurological findings were observed after the convulsions disappeared. Electroencephalography also revealed no abnormalities. Computed tomography (CT) showed a hyperdense area in the...
left precentral gyrus, which was enhanced by contrast medium (Fig. 2). $T_1$-weighted magnetic resonance (MR) imaging demonstrated a heterogeneously intense lesion, which was enhanced by gadolinium-diethylenetriaminepenta-acetic acid, and a hypointense rim, which suggested a hemosiderin ring (Fig. 2). $T_2$-weighted MR imaging also disclosed an intense mosaic mass and a hypointense rim. Cerebral angiography did not show the enhanced mass, but revealed caput Medusae-like, abnormal blood vessels in the perifocal region and draining into the dilated abnormal cortical vein, suggestive of venous angioma (Fig. 3).

A left frontotemporal craniotomy was performed. The central sulcus was separated from the posterior abnormal vein, which angiography had suggested as draining the venous angioma, to reveal xanthochromic cortex. Corticotomy at this site demonstrated a dark red abnormal vascular mass, from which blood was draining into the abnormal vein. The vascular mass was totally extirpated without damaging the surrounding abnormal blood vessels. Histological examination of the mass revealed an aggregate of blood vessels of various sizes, in which large amounts of hemosiderin deposits were present (Fig. 4). No nerve tissue was observed between the blood vessels. The vascular vessel walls varied from thin to thick, but no elastic fibers were seen. Postoperative MR imaging showed that the venous angioma was preserved without damage (Fig. 5). The patient was discharged from the hospital one week after the operation with no neurological deficits. Administration of anticonvulsant was discontinued 2 years after the operation, and no convulsive seizure occurred in the 2 years.

**Case 2:** A 28-year-old right-handed female was admitted to our hospital suffering from convulsions. No abnormal neurological findings were observed after the convulsion disappeared. Electroencephalography also showed no abnormalities. CT showed a hyperdense area, which was enhanced by contrast medium accompanied by perifocal, abnormally...
thick, linear enhancement (Fig. 6 left). Angiography revealed caput Medusae-like findings (Fig. 6 center). MR imaging revealed a lesion with the same appearance as in Case 1 in the right inferior frontal gyrus (Fig. 6 right).

A right frontotemporal craniotomy was performed. During the operation, xanthochromic cortex was found by separating the sulci along the abnormal veins, as in Case 1. Corticotomy of a few millimeters at this site demonstrated an abnormal vascular mass accompanied by surrounding hematoma. This lesion including the surrounding gliosis was totally resected. A thick vein, apparently a vein of venous angiomatosis, was connected to this vascular mass by a thin vein, but the venous angioma was not damaged. During the operation, the spike waves disappeared on the corticogram following resection of the lesion (Fig. 7).

Histological examination showed the abnormal
vascular mass was a cavernous angioma. Postoperative cerebral angiography showed the venous angioma was intact. The patient was discharged from the hospital one week later with no symptoms. Subsequent treatment has not included an anticonvulsant, but the patient has had no convulsions for the past 2 years.

Discussion

The association between cavernous angioma and venous angioma is well-known. It has been suggested that every case of venous angioma is complicated by cavernous angioma, but this requires a close radiological search to identify in every case. Only about 50 cases of the coexistence of cavernous angioma and venous angioma have actually been proved. Eight of these 50 cases had two angiomas in the same region and operative findings which were clearly described, although no definitive explanation for the coexistence of these lesions has been given. The explanation most address the following facts: the frequency is too high to conclude that the coexistence is incidental; the two angiomas occur in the same region; and intraoperative findings reveal a draining vein leading from the cavernous angioma to the venous angioma. However, whether the colocalization is caused by a common pathogenesis due to developmental abnormality or a hemodynamic problem is unclear. Venous angioma may be a condition of venous hypertension, resulting in the formation of cavernous angioma, and abnormal enhancement on MR imaging may be evidence of venous hypertension. No imaging in the present cases suggested venous hypertension. The lesion thought to be attributable to common pathogenesis rather than venous hypertension, as occurrence of cavernous angioma was not located at the distal end of the draining vein, although the pressure on the central side of the draining vein was probably lower than on the peripheral side.

Surgical treatment of the patients was intended to remove the epileptogenicity, to prevent future bleeding, and to make a definitive histological diagnosis. Both patients were asymptomatic for 2 years following discontinuation of anticonvulsant therapy. Disappearance of spike waves was observed in Case 2 during the operation. Treatment to prevent bleeding for coexisting cavernous angioma and venous angioma should be approached differently to that required for each individual condition, because bleeding of the cavernous angioma makes preservation of the venous angioma difficult. In the present cases, the cavernous angioma was connected to the draining vein of the venous angioma by a thin vein, and the angiomas were in very close proximity. Therefore, hematoma due to bleeding from the cavernous angioma will damage both the medullary vein and the abnormally thick draining vein of the venous angioma. Since this thick abnormal draining vein of the venous angioma also drains the normal medullary vein, swelling of the brain due to compression by the hematoma or surgical injury can be expected. Therefore, if this coexistence is diagnosed, surgery to remove the cavernous angioma should be performed before bleeding occurs. Of course, if bleeding from the cavernous angioma occurs, extirpation should be performed, and the venous angioma left untouched. The annual bleeding rate of cavernous angioma is not more than 1%, and the annual incidence of convulsion is 1.51%, indicating that symptoms are not necessarily frequent. However, when cavernous angioma is extirpated before bleeding, the incidence falls an extremely low level. In addition, the onset of symptoms in eight cases of the coexistence of cavernous angioma and venous angioma may have been associated with bleeding into the cavernous angioma. Thus, cavernous angioma should be positively extirpated even if located in an eloquent area as in our Case 1.

Patients with cavernous angioma manifesting as convulsions can be cured by surgical extirpation of
the angioma at the time of detection to prevent future bleeding. When the cavernous angioma is complicated by venous angioma, as in the present cases, early extirpation is preferred.

References


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