[Case Report]

Removal of Double Cavernous Angioma of the Frontal Lobe using a Three-Dimensional Printed Model: A Case Report

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Abstract **:** Cerebral cavernous angiomas are vascular anomalies with dilated spaces. We report the case of rare double cavernous angiomas causing higher brain dysfunction. A 74-year-old man exhibited cognitive dysfunction. Magnetic resonance imaging showed two tumors with hemorrhage in the left frontal lobe. Preoperative diagnosis was hemorrhage caused by cavernous angiomas. A 3D model of the double cavernous angioma was made to confirm their association with cortical veins and tumors. Tumors were removed using a single small corticotomy. This is the first report of a rare double cavernous angioma and the 3D printed model facilitated removal of the tumors.

Keywords **:** cavernous angioma, frontal lobe, hemorrhage, magnetic resonance imaging, three-dimensional printer.

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Introduction

Cavernous angiomas, also known as cavernous malformations or cavernomas, are vascular malformations characterized by the presence of sinusoid-like capillary vessels [1−4]. Symptoms of cavernous angiomas are typically seizures or focal deficits caused by a single tumor [5]. The hemorrhage rate reportedly tends to be low [5−8].

Here, we describe a patient who exhibited higher brain dysfunction owing to simultaneous hemorrhage of double cavernous angiomas. Double cavernous angioma in the same region and causing higher brain dysfunction is extremely rare. We excised them using a three-dimensional (3D) printed model. Recently, 3D models have been used for preoperative simulation, especially in bone or vascular surgeries. We used it for tumor removal. The 3D model facilitated the understanding of the relationship of the double tumor. To the best of our knowledge, this is the first report of a rare double cavernous angioma treated using a 3D printed model.

The patient provided informed consent for publication of his clinical data and images.

Case Report

A 74-year-old man was admitted to our department because of mild cognitive impairment. His symptoms began with inability to drive, caused by sudden worsening of cognitive impairment. He had been prescribed direct oral anticoagulants for arterial fibrillation. Upon admission, neurological examination showed mild right hemiparesis (manual muscle test 4/5) and cognitive impairment. The mini-mental state examination

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(MMSE) score was 18/30, and the frontal assessment battery (FAB) score was 14/18. Computed tomography (CT) showed two hemorrhagic masses in the left frontal lobe (Fig. 1A). These masses were 3 cm and 1.5 cm in size, with perifocal edema. Magnetic resonance imaging (MRI) showed two separate masses exhibiting different intensities. They demonstrated a "popcorn" appearance on fluid-attenuated inversion recovery (Fig. 1B,C). Contrast-enhanced T1-weighted images did not show any enhancement of these masses. The patient underwent cerebral angiography, which did not reveal any tumor blush. The suspected preoperative diagnosis was cavernous angioma with hemorrhage. We chose to remove these tumors simultaneously. To achieve minimal invasion, we planned removal of the small tumor on the top followed by removal of the large tumor using the same corticotomy. However, we presumed that it would be difficult to approach the large tumor from the point of removal of the small one. Therefore, we made a 3D model with the cortical vein and confirmed the distance and direction from the small tumor to the large one.

3D-printing procedure

We used the standard DICOM image files of the MRI contrast-enhanced T1-weighted image to obtain the stereolithography (stl) file format required for 3D printing. The DICOM images were processed using the OsiriX 32-bit software (Pixmeo SARL, Bernex, Switzerland) to obtain the images of these tumors with the cortical vein in the stl file format. This image in stl file format was printed using the UP Plus2 3D printer (Delta-Microfactory, China) with a 1.75 mm acrylonitrile-butadiene-styrene resin filament. Fig. 2 shows the image of tumor with cortical vein and the 3D printed model.

Under the monitoring of the motor-evoked potential and neuro-navigation, the patient underwent left frontal craniotomy, which was performed from the top of the small tumor by confirming the position of the nearest cortical vein using the 3D model (Fig. 3A). The lesion contained old liquefied and solid hematoma parts. The solid part was firm and surrounded by gliotic changes, and it could be excised completely. The pathological diagnosis was confirmed as cavernous angioma. Normal intact white matter was present between the two masses and confirmed the direction and distance from the small to large tumor using the 3D model (Fig. 3B, C); the large tumor was revealed when the white matter was removed (Fig. 3D). The large tumor was separate and exhibited the same features as the small one. It contained an old hematoma; the capsule was removed by gradual excision from the same corticotomy area. Complete removal was achieved. Histopathological analysis of the resected lesion revealed vascular spaces containing blood elements, lined by a single layer of endothelial cells; the lesion

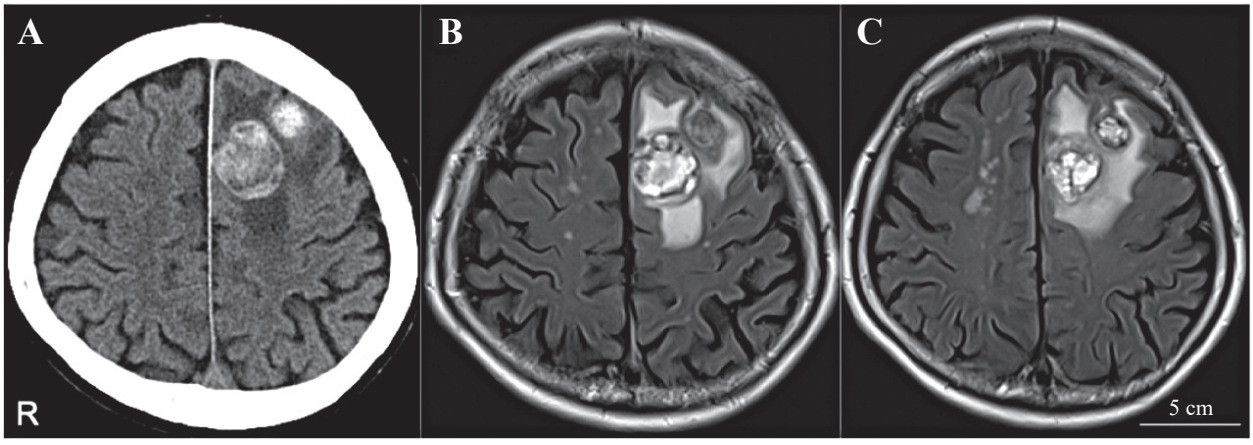

Fig. 1. Double hemorrhages in the left frontal lobe. A: Computed tomography (CT) shows two hemorrhages in the left frontal lobe. B, C: Magnetic resonance imaging (MRI) shows two tumors with different intensities and popcorn appearance on fluid-attenuated inversion recovery.

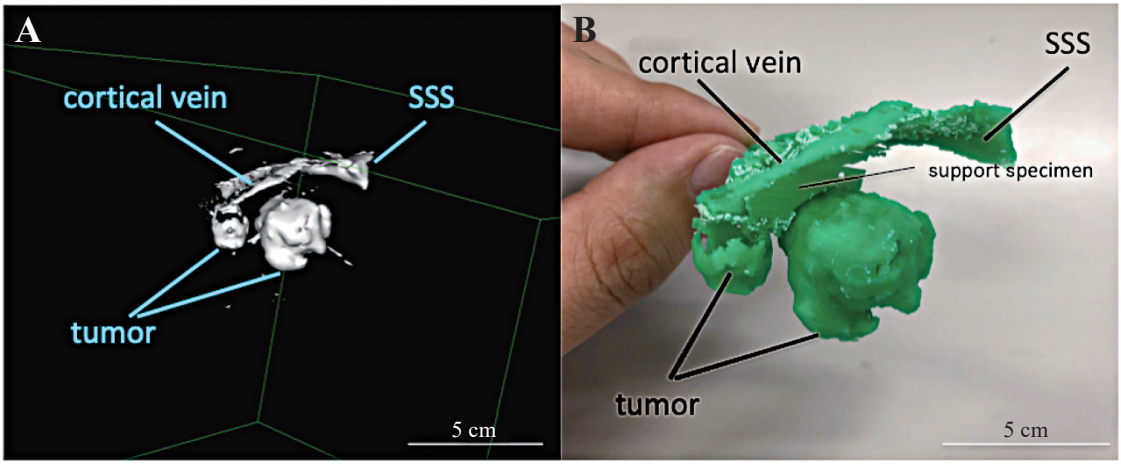

Fig. 2. Preparation for 3D model. A: Image of tumors and veins. B: 3D model of tumors and cortical vein and sagittal straight sinus. The 3D models include a support specimen.

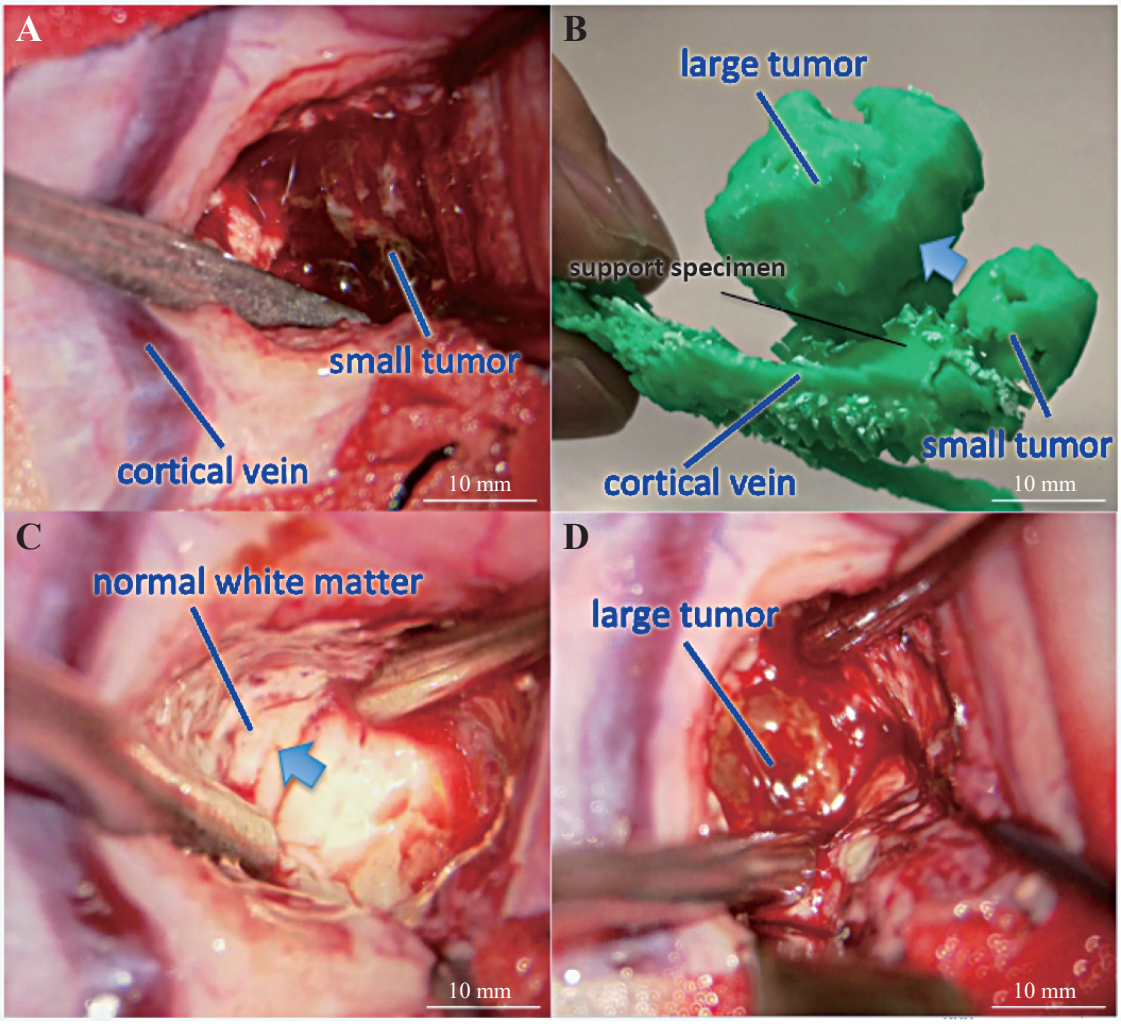

Fig. 3. Surgical procedure. A: Corticotomy is performed and the small tumor is found. B: Direction from the small tumor is confirmed. Arrow shows the direction to the large tumor. C: After removing the small tumor, normal white matter is sucked to direction point. D: Large tumor is found.

was confirmed as cavernous angioma. One week after the operation, the patient showed improvement in neurological deficit; MMSE and FAB improved to 29 and 18, respectively. The patient could resume driving.

Discussion

Cavernous angiomas comprise approximately 5−13% of all cerebral vascular malformations [1,9]. Patients with cavernous angiomas typically remain asymptomatic. Most symptomatic patients present with seizure or focal neurological deficit. In the present case, the patient exhibited higher brain dysfunction. The two cavernous angiomas that caused the symptoms were located at the same site and exhibited concomitant hemorrhage; this was an extremely rare form of cavernous angioma that has not been reported.

Three-dimensional printing was introduced approximately a decade ago. The cost of 3D printers has recently decreased, and our 3D printer cost approximately US$ 1,000. In medicine, 3D printing technology has been used to create personalized models of the patient's anatomy for surgical training as well as for the creation of customized prostheses and implantable medical devices [10−12]. In the present case, we could prepare a 3D model of the tumor at a low cost within 3 hours. Thus, we could understand not only the tumor, but also its relation to the surrounding tissues.

In our case, to remove two tumors from the same corticotomy area, we needed to know the relationship of the two tumors prior to the operation. Using navigation was useful; however, it may not have been accurate because of shifting during the operation. The 3D model provided us information regarding the distance and direction of the two tumors by allowing us to hold it in our hands, leading to easier identification of the tumors. The 3D model also provided information regarding the corticotomy area by confirming the location of the nearest vein. The corticotomy area was confirmed by neuro-navigation; however, it was more accurately determined using the 3D printed model.

Thus, we found that the 3D model was useful for removal of the tumor using a single corticotomy in the present case. It was also useful to determine the corticotomy area.

Conclusion

Double cavernous angiomas in the same location are extremely rare. They cause sudden dysfunction of the higher brain by hemorrhage. In the present case, they were easily removed with minimum invasion by using the 3D printed model. The 3D printer is useful owing to its low cost, ability to provide a model in a short time, and ability to provide accurate details to facilitate surgical excision.

Conflicts of Interest

The authors declare no conflicts of interest.

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三次元モデルを用いて摘出術を行った二箇所の前頭葉海綿状血管腫

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要　　旨：脳実質内の海綿状血管腫は拡張した腔を持つ血管奇形である．我々は，二箇所の海綿状血管腫が高次脳機能障害を呈した稀な症例を経験したため報告する．症例は74歳男性で認知機能障害を呈していた．MRI検査にて左前頭葉に出血を伴う二箇所の腫瘍性病変を認め，術前診断では海綿状血管腫の出血と診断された．腫瘍と皮質静脈との関連を調べるために，この腫瘍に対して三次元モデルを作成した．二つの腫瘍の関連を把握することで二箇所の腫瘍は小さな皮質切開から摘出を行うことができた．今回の症例は，二箇所隣接して存在する稀な海綿状血管腫であり，三次元モデルが二箇所の腫瘍を摘出するのに有用であった．

キーワード：海綿状血管腫，前頭葉，出血，MRI，三次元モデル．

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