

Cavernous Hemangioma in the Middle Cranial Fossa & Cavernous Sinus

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Extracerebral cavernous hemangiomas are rare vascular tumors that are very difficult to remove because of severe intraoperative bleeding. We report a case of 57-year-old male with extracerebral cavernous hemangioma with review of 126 cases in the literature. Patient presented with blurred vision, diplopia, numbness on the left side of his face. Magnetic resonance imaging revealed a well defined mass of 3×4×3cm size with heterogenous iso-or hypointensity on T1-weighted image showing strong homogeneous contrast enhancement and marked hyperintensity on T2-weighted image. Digital subtraction angiography(DSA) revealed a faint tumor blush by feeders from the left internal carotid artery(ICA) and left external carotid artery(ECA) in the delayed phase. Even with profuse intratumoral bleeding, near total removal was achieved. In addition to preoperative neurologic deficits such as ophthalmoplegia, facial numbness in the V1-2 dermatomes, ptosis appeared postoperatively.

KEY WORDS : Extracerebral cavernous hemangioma · Cavernous sinus · Middle cranial fossa · High signal intensity on T2WI.

Introduction

Cavernous hemangioma is one of four most common types of cerebral vascular malformation⁴⁾. Extracerebral cavernous hemangiomas are rare vascular tumors, only accounting for 0.4 to 2% of the intracranial cavernous hemangiomas, and most commonly located in the cavernous sinus or middle cranial fossa¹⁾. In these sites, differential diagnosis with tumors such as meningioma or schwannoma is often

difficult⁵⁾. It has long been a great challenge to neurosurgeons because of the difficulties in preoperative diagnosis and surgical removal of tumor without excessive hemorrhage⁶⁾. In this report, we present our experience.

Case Report

A 57-year-old male presented with blurred vision, diplopia, numbness on the left side of his face. He had a vis-

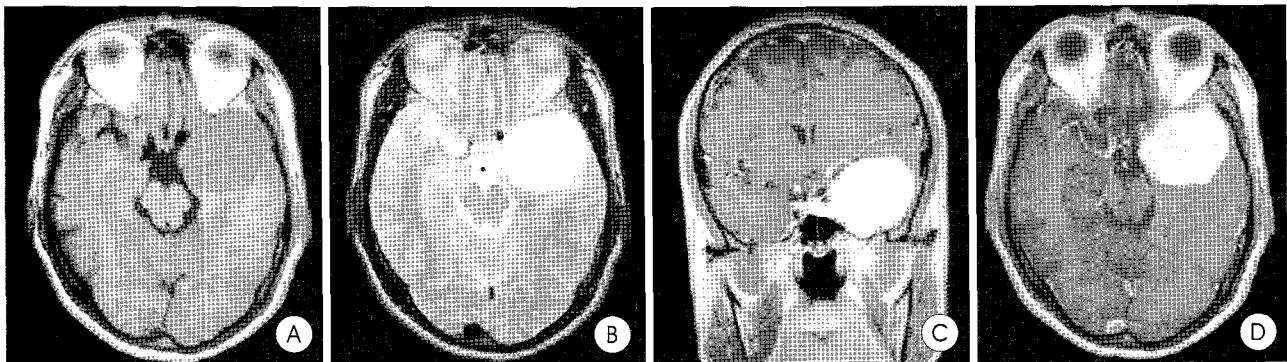


Fig. 1. Preoperative magnetic resonance image(MRI) (A : T1WI, B : T2 WI, C : Coronal image with enhancement, D : Axial image with enhancement) MRI revealing a large tumor in the left cavernous sinus, extending into the middle cranial fossa, with bright enhancement and a well defined margin. T2-weighted image showing that the tumor exhibited marked hyperintensity that is similar to that of cerebrospinal fluid.

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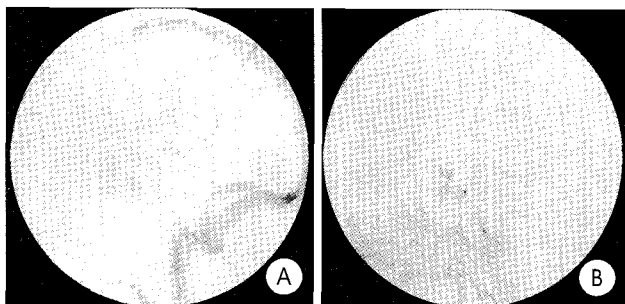


Fig. 2. Tumor stain is seen in the venous phase (A : Left internal carotid angiography. B : Left external carotid angiography).

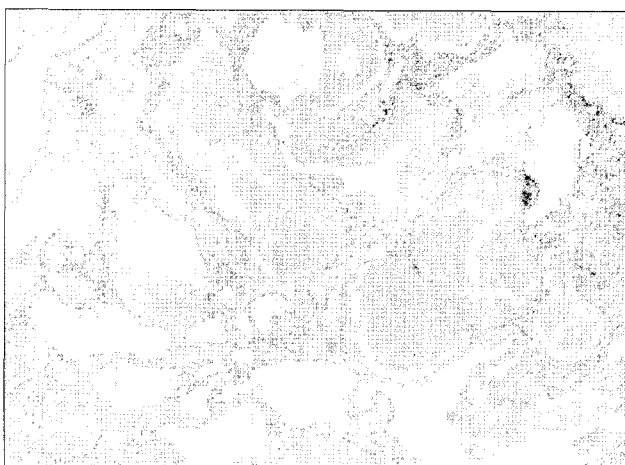


Fig. 3. The histology of the tumor is composed of dilated blood containing channels lined with flattened endothelium separated by fibrous tissue (H&E, $\times 100$).



Fig. 4. Magnetic resonance image obtained 9 month after surgery, showing no residual tumor.

ual acuity of (1.2/0.5). Neurological examination revealed left side sixth nerve palsy and hypesthesia in the left V1-3 dermatomes. Magnetic Resonance Imaging(MRI) revealed a well defined mass of $3 \times 4 \times 3$ cm size in the left cavernous sinus, extending to the middle cranial fossa with heterogenous iso-or hypointensity on T1- weighted image showing strong homogenous contrast enhancement and marked hyperintensity on T2-weighted image (Fig. 1). Digital subtraction angiography(DSA) revealed a faint tumor blush by feeders from the left internal carotid artery(ICA) and left external carotid artery(ECA) in the delayed phase (Fig. 2). Tumor embolization was tried but failed. Preoperative differential diagnoses were meningioma,

cavernous hemangioma. Left pterional craniotomy was performed. The tumor was adherent to the 3rd, 5th, 6th cranial nerve and the lateral wall of the left cavernous sinus. Resection of the tumor led to profuse bleeding. Near total tumor resection could be achieved. Microscopic examination of the specimen confirmed the diagnosis of cavernous hemangioma (Fig. 3). Immediate postoperative neurological examination revealed new 3rd nerve palsy. The patient had 4 seizure attacks after operation. Radiotherapy started for remnant tumor at the 28th day after operation. Conventional fractionated radiation of 60Gy to tumor was given for 6.5weeks. Five months postoperatively, ptosis, ophthalmoplegia, facial numbness in the V1-2 dermatomes remained. But there was no more seizure attack. His visual acuity was (1.2/0.2). Nine month after surgery, follow up MRI revealed no residual tumor (Fig. 4).

Discussion

Among the 140 cases of extracerebral cavernous hemangioma in the literature, the data of 126 cases were available for analysis and review. Summary of the cases are listed on the Table 1.

The patients were aged from 14 to 79 years with peak incidence at the 5th decades. There were 27 males and 100 females with a female/male ratio of 3.5:1. There were a predominance of orientals with a oriental/westerner ratio of 2:1.

Initial presenting symptoms are listed on the Table 2. Generally, extracerebral cavernous hemangioma occurred insidiously and patients deteriorated gradually. The duration between occurrence of symptoms and the admission ranged from 1day to 10years, with an average of 20 months. In contrast to the most cerebral cavernous hemangiomas, bleeding has been very rarely reported in the extracerebral cases⁵. But Escott et al. reported a case of suprasellar cavernous malformation presenting with extensive subarachnoid hemorrhage². Important characteristic of these lesions is that the clinical symptoms may be exacerbated during pregnancy and exhibit spontaneous remission after delivery¹³.

The tumors are generally isodense to hyperdense on noncontrast computed tomography(CT) with strong contrast enhancement^{5,6}. MRI reveals well enhancing masses on contrast enhancement that are hypointense or isointense in T1-weighted image and markedly hyperintense in T2-weighted image¹³. From the angiographic findings of 81 cases, tumor blush was positive in 52 cases (64%). But no stain has been reported in 29 cases. Embolizations were performed in 5 cases. But the results were not successful.

Cavernous hemangiomas are surgically curable, however, total removal has been very difficult due to excessive bleeding^{11,12,15}.

Table 1. Summary of 127 cases with Intracranial extracerebral cavernous hemangioma

Author(year)	No. of case	Author(year)	No. of case	Author(year)	No. of case
Watanabe et al.(1943)	1	Min et al.(1990)	2	Seo et al.(2000)	1
Kawaguchi et al.(1965)	1	Meyer et al.(1990)	8	Thompson et al.(2000)	4
Kajimo et al.(1972)	1	Yamamoto et al.(1992)	1	Kida et al.(2001)	3
Ogasawara et al.(1973)	1	Linskey et al.(1992)	3	Escott et al.(2001)	1
Itoh et al.(1977)	1	Perry et al.(1993)	1	Tannouri et al.(2001)	1
Ichozaki et al.(1980)	2	Lee JG et al.(1993)	1	Paladino et al.(2001)	1
Kim TS et al.(1980)	1	Lee JK et al.(1993)	1	Cobbs et al.(2001)	1
Waga et al.(1981)	1	Goto et al.(1993)	1	Bondi et al.(2002)	5
Fang et al.(1982)	8	Maruishi et al.(1994)	1	Kim IM et al.(2002)	1
Namba et al.(1983)	1	Lombardi et al.(1994)	4	Kocak et al.(2002)	1
Rosenblum et al.(1986)	1	Suzuki et al.(1996)	7	Shakh et al.(2002)	1
Shibata et al.(1987)	3	Gupta et al.(1998)	1	Zhou et al.(2003)	20
Katoh et al.(1987)	3	Ohata et al.(1999)	5	Peker et al.(2004)	5
Kudo et al.(1989)	1	Shi et al.(1999)	10	Puca et al.(2004)	1
Sawamura et al.(1990)	1	Gliemroth et al.(2000)	1	present case	1
Rigamonti et al.(1990)	3	Tu et al.(2000)	3		

Table 2. Initial presenting symptoms (N=127)

Symptoms	No. of case
Decrease in visual acuity	48
Visual field defect	12
EOM limitation or diplopia	78
Exophthalmos	13
Ocular pain	10
Headache	55
Facial numbness	41
Facial palsy	5
Amenorrhea or galactorrhea	13
Obesity	5
Aggravation during pregnancy	9
Seizure attack	2
Altered mentality	3

Table 3. Treatment modalities used on patients

	OP(+RT)	Non op
Total removal	53	Radiosurgery 5
Subtotal removal	24	
Partial removal	34	Preop death 1
Biopsy	10	
Total(127)	121(29)	6

Table 4. Clinical response after therapy

	Improved	Aggravated	No change	Dead	Total
Patients	75	23	19	10	127

Table 5. Causes of operative morbidity (N=106)

Morbidity	Patients
Ophthalmoplegia	39
Blindness	7
5th CN deficit	17
7th CN deficit	1
Hemiparesis or aphasia	4

To minimize blood loss during resection of these hemorrhagic tumors, various surgical methods that decrease tumor size and vascularity have been reported^{9,10,14}. Some authors have controlled troublesome bleeding from cavernous hemangiomas which is not usually controlled by bipolar coagulation by packing the sites of bleeding with a fibrin glue-soaked oxidized cellulose or gelatin sponge^{7,15}. Because of the difficulty of total tumor removal, radiotherapy was chosen as an alternative method before and/or after surgery^{3,14}. Pecker et al. reviewed radiosurgically treated cavernous sinus cavernous hemangiomas. Radiologically near complete response was found in 4 out of

16 patients. None of the patients demonstrated tumor growth after radiosurgery. Neurological status improved in 9 patients. No complication was seen in a total of 16 patients⁸. Operation were performed in 121 cases. Among them, 29 patients underwent combined radiotherapy. Five patients underwent only radiosurgery without surgery. Total removal was achieved in 53 patients. Subtotal removal, partial removal and biopsy were achieved in 24, 34, 10 patients, respectively (Table 3). Among the 127 patients, 75 patients improved in their neurological symptoms and signs, 19 no change and 33 aggravated or dead after therapy (Table 4).

Histologically, the hemangioma includes endothelium-lined, thin walled vessels. Shi et al. described two types of hemangiomas, those with a large number of thin walled sinusoids and endothelium-lined capillaries and a second type with well formed vasculature and an increased amount of connective tissue. The first type was more prone to bleed excessively than the second type and less likely to be removed completely¹³. Our case belonged to the first type.

In 106 cases, a detail of postoperative morbidity was described. Most common operative morbidities were ophthalmoplegia and the 5th CN deficit (Table 5).

Conclusion

Extracerebral cavernous hemangiomas usually present with mass effect including cranial nerve deficits and headache. Specific MR findings, such as strong homogenous contrast enhancement and marked hyperintensity on T2-weighted image, are helpful for diagnosis. We experienced a case of ex-

tracerebral cavernous hemangioma, which located in the cavernous sinus and middle cranial fossa, and reported its clinical features with review of literatures.

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