

## CASE REPORT

# Trigonal cavernous malformation with intraventricular hemorrhage : A case report and literature review

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**Abstract :** We reported a case of trigonal cavernous malformation (CM) with intraventricular hemorrhage. This 67-year-old woman experienced sudden onset of loss of consciousness and her Glasgow Coma Scale (GCS) was 5 points (E1V1M3) on admission. CT scan demonstrated intraventricular hemorrhage and acute hydrocephalus. Angiography did not demonstrate any vascular abnormality. Ventricular drainage was performed for acute hydrocephalus and the postoperative course was good. CT showed a hyperdense lesion in the left trigone, which was contrast-enhanced on T1-weighted MR. Removal of CM was performed via the left middle temporal sulcus. We conducted a Pub Med search for trigonal CM and found 17 cases. Herein we discuss the symptoms, CT and MR findings and treatment. *J. Med. Invest.* 59 : 275-279, August, 2012

**Keywords :** cavernous malformation, hydrocephalus, intraventricular hemorrhage, trigone

## INTRODUCTION

Intraventricular cavernous malformation (CM) is rare, comprising only 2.5-10.3% of patients with cerebral CMs (1, 2). Of intraventricular CM, only about 20% are located in the trigone of the lateral ventricle (3). The first report of trigonal CM was published in 1977 by Coin (4). Based on our search of PubMed, trigonal CM has been reported in 17 cases over about 30 years. We encountered a rare case of trigonal CM and discuss the symptoms, CT and MR findings and the treatment.

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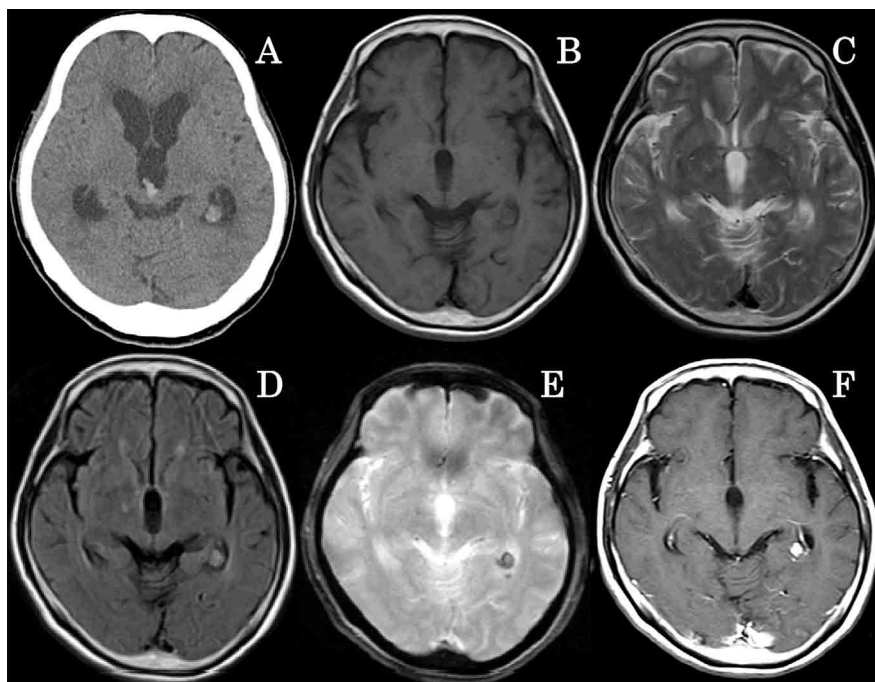
Received for publication March 29, 2012 ; accepted May 18, 2012.

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## CASE REPORT

### *Examination and preoperative course*

A 67-year-old right-handed woman with a history of hypertension, experienced a sudden onset of consciousness disturbance. She was immediately transferred to our hospital. On admission, her GCS (Glasgow Coma Scale) was 5 points (E1V1M3), but there was no paresis. The pupils were equal in size and reacted to light. Corneal reflexes were present bilaterally. CT scan showed an intraventricular hemorrhage in the left trigone, in the cerebral aqueduct and acute hydrocephalus (Figure 1). Angiography did not show any vascular abnormality. We performed ventricular drainage. Seven days after surgery, her condition was good. MR imaging showed a lesion measuring 10 mm within the trigone of the left lateral ventricle. It was isointense on T1-weighted, hypointense on T2-weighted, hyperintense on FLAIR imaging and hypointense on T2\*



**Figure 1** Admission CT scan showing a hyperdense lesion in the trigone of the left lateral ventricle, and a small amount of hemorrhage in the cerebral aqueduct along with acute hydrocephalus (A). The lesion was isointense on T1-weighted (B) ; hypointense on T2-weighted (C) ; hyperintense on FLAIR (D) ; and hypointense on T2\* MR (E). Contrast-enhanced MR image showing Gd-DTPA enhancement (F).

imaging and well enhanced with gadolinium on T1-weighted imaging (Figure 1).

### *Surgery*

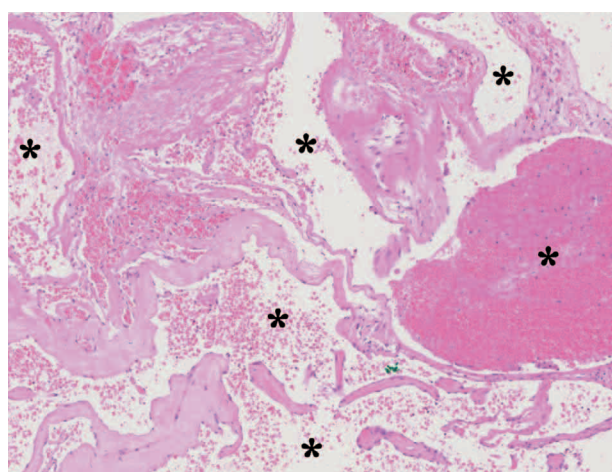
The patient underwent total removal of the mass via the left middle temporal sulcus. The middle temporal sulcus was incised and the left ventricle opened. The choroidal plexus was hypertrophic, red, and led to the trigonal lesion. The mass was bluish-red and packed with thin-walled vessels. A draining vein was seen leading out of the lesion to the inner surface of the lateral ventricle. The mass was totally removed.

### *Postoperative course*

The patient transiently demonstrated mild dyslexia and mild acalculia, with no visual field defects. She did not show any neurological findings 3 weeks later. Postoperative MR imaging confirmed that the lesion in the trigone of the left lateral ventricle had been removed.

### *Pathological Findings*

Histologically, the lesion was composed of dilated vessels filled with blood. The vascular walls were lined by endothelial cells with fibrous stroma. There was no glial tissue between the vessels. The pathological diagnosis was CM (Figure 2).



**Figure 2** Dilated vessels (\*) lined by endothelial cells with fibrous stroma. There was no glial tissue between the vessels. H & E,  $\times 100$

## DISCUSSION

Intraventricular CMs occur in only 2.5-10.3% of patients with cerebral CM (1, 2). Trigonal CM is even rarer. Among the 17 cases of trigonal CM that we have collected, the median patient age was 36.1 years and there were eight males and nine females (Table). Lesion size was reported to be around 1 cm or larger. Our case was a so-called microangioma (5).

Table

Case	Year	Author	Reference	Age	Sex	Symptoms	Side	Tumor size (cm)	Angiography	CT scan	Surgery	Outcome
1	1977	Coin <i>et al.</i>	4	36	F	Seizures	Rt	NA	normal	hyperdensity	NA	Hemianopia
2	1977	Numaguchi <i>et al.</i>	22	43	M	Mass lesion	Rt	NA	venous stain and venous pooling	NA	NA	Hemiplegia, hemianopia
3	1979	Pau and Orynesu <i>et al.</i>	23	56	F	IVH	Lt	NA	not recognizable	NA	inoperable	Dead
4	1983	Iwasa <i>et al.</i>	24	8	F	Mass lesion	Lt	3	NA	hyperdensity	temporal craniotomy	Improvement
5	1983	Handa <i>et al.</i>	7	73	M	Mass lesion	Rt	3	no tumor stain	NA	lower posterior parietal lobe incision	Improvement
6	1986	Yamasaki <i>et al.</i>	25	73	M	Mass lesion	Rt	3	no feeding artery, tumor stain, draining vein	hyperdensity, calcification	NA	Improvement
7	1989	Andoh <i>et al.</i>	16	62	F	Mass lesion	Lt	3	no revealed homogenous overshadowing	hyperdensity	middle temporal gyrus	Dead
8	1990	Tatagiba <i>et al.</i>	7	35	M	Seizures	Rt	4	normal	hyperdensity	calcarine fissure	Improvement
9			24	M	Mass lesion	Lt	3	draining vein	hyperdensity	transcortotomy (behind angular gyrus)		Improvement
10	1993	Miyagi <i>et al.</i>	17	3	F	Mass lesion	Lt	NA	no abnormal staining	hyperdensity	transsylvian transventricular	Improvement
11	2003	Nieto <i>et al.</i>	26	11	F	Seizures	Lt	5	NA	NA	temporal craniotomy	Improvement
12	2006	Kumar <i>et al.</i>	15	8	M	Mass lesion	Rt	5	NA	brilliant enhancement	inferior temporal, superior temporal sulcus	Improvement
13			19	F	Mass lesion	Lt	5	no tumor blush or evidence of AVM	NA	NA	middle temporal gyrus	Hemiparesis
14			20	M	Mass lesion	Rt	5	no abnormality	NA	NA	middle temporal gyrus	Improvement
15	2007	Gonzalez-Darder	27	25	M	IVH	Rt	3	not pathological	hyperdensity	trans-sulcus temporal posterior	Improvement
16	2008	Stavrinou <i>et al.</i>	18	52	F	Mass lesion	Rt	3	unremarkable	NA	low postcentral transcortical	Improvement
17	2012	Ohbuchi <i>et al.</i>		67	F	IVH	Lt	1	no vessel malformation	hyperdensity	middle temporal gyrus	Improvement

IVH : intraventricular hemorrhage, NA : not available, F : female, M : male, Rt : right, Lt : left

The first symptom was seizure in three cases (17.6%), intraventricular hemorrhage in three (17.6%) and mass lesion in eleven (64.7%) (Table). The symptoms of mass lesion showed a higher incidence than seizure or intraventricular hemorrhages in trigonal CMs. Katayama *et al.* reported that the reason for the higher incidence of the mass effect may be direct compression of vital structures. Cavernous malformation is often voluminous as it grows in a preexisting cavity (6).

Among 138 cases of intracranial symptomatic CMs reviewed by Simard *et al.* 40 (29%) presented with extralesional hemorrhage. A similar incidence (27%) of subarachnoid and/or intraventricular hemorrhage was reported in intraventricular CMs (7). Among the 77 cases of intraventricular cerebral CMs reviewed by Kivelev *et al.*, 11 (14%) presented with

intraventricular hemorrhage and intraventricular CMs had a higher tendency for rebleeding than CMs in other locations (8, 9). It is expected that our case showing intraventricular hemorrhage would also have a higher tendency to rebleed ; therefore, we selected surgical removal.

Typical CT findings of intraventricular CM include moderately hyperdense intraventricular nodular lesions showing mild contrast enhancement and moderate signs of a mass effect (10). The CT findings in 9 cases of trigonal CM that reported CT density indicated that the lesion was hyperdense. Typical findings of CMs on MRI are : central area of hypersignal, corresponding to the presence of methemoglobin, associated with areas of hyposignal due to calcification and extense fibrosis within the lesion on T1 and T2 weighted images ; marked enhancement ;

peripheral hypointense area due to the presence of hemosiderin ; moderate mass effect compared to the size of lesion ; and slight perifocal edema (10). Cerebral angiography rarely shows abnormalities other than a nonspecific avascular mass (9, 11, 12, 13, 14). Angiography plays a role in excluding AVM (15).

Differential diagnoses are choroid plexus papilloma, teratoma, astrocytoma, ependymoma, neurocytoma, metastasis, meningioma and arteriovenous malformation (6, 16, 17, 18).

The surgical approach for trigonal lesions depends on the size of the lesion and whether the lesion is in the dominant hemisphere. We choose the transtemporal approach to the lesion even in dominant hemispheres. This approach has many theoretical advantages : it is close to the tumor ; the anterior choroidal artery can be managed early (19, 20) ; and sufficient decompression and a wide operative field are obtainable because the inferior horn of the lateral ventricle is generally dilated. The disadvantages are (21) : homonymous hemianopsia may occur at least slightly, even though the approach is made parallel to the optic radiation ; individual differences have been noted at the site of Wernicke's area, so fluent aphasia might occur even when there is no invasion in the posterior 1/3 of the superior temporal gyrus on the dominant side ; and tumors that develop on the posterior body are hardly removable (16). In the literature, twelve cases (70%) showed improvement. Our patient initially demonstrated mild dyslexia and mild acalculia, which completely disappeared within 3 weeks.

## DISCLOSURE

There is no COI status to disclose. Hidenori Ohbuchi  
 There is no COI status to disclose. Yasuhiko Osaka  
 There is no COI status to disclose. Takahiro Ogawa  
 There is no COI status to disclose. Masataka Nanto  
 There is no COI status to disclose. Yoshikazu Nakahara  
 There is no COI status to disclose. Kanade Katsura  
 There is no COI status to disclose. Hiroshi Tenjin  
 There is no COI status to disclose. Hidetoshi Kasuya

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