CASE REPORT

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

Hidenori Ohbuchi[1)3)], Yasuhiko Osaka[1)], Takahiro Ogawa[1)], Masataka Nanto[1)], Yoshikazu Nakahara[1)], Kanade Katsura[2)], Hiroshi Tenjin[1)], and Hidetoshi Kasuya[3)]

[1)]Department of Neurosurgery, [2)]Department of Pathology, Kyoto Second Red Cross Hospital, Kyoto, Japan ; and [3)]Department of Neurosurgery, Tokyo Women's Medical University Medical Center East, Tokyo, Japan

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.

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

Address correspondence and reprint requests to Hidetoshi Kasuya, Department of Neurosurgery, Tokyo Women's Medical University Medical Center East, 2-1-10 Nishiogu, Arakawa-ku, Tokyo 116-8567, Japan and Fax : +81-3-3819-8680.

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*

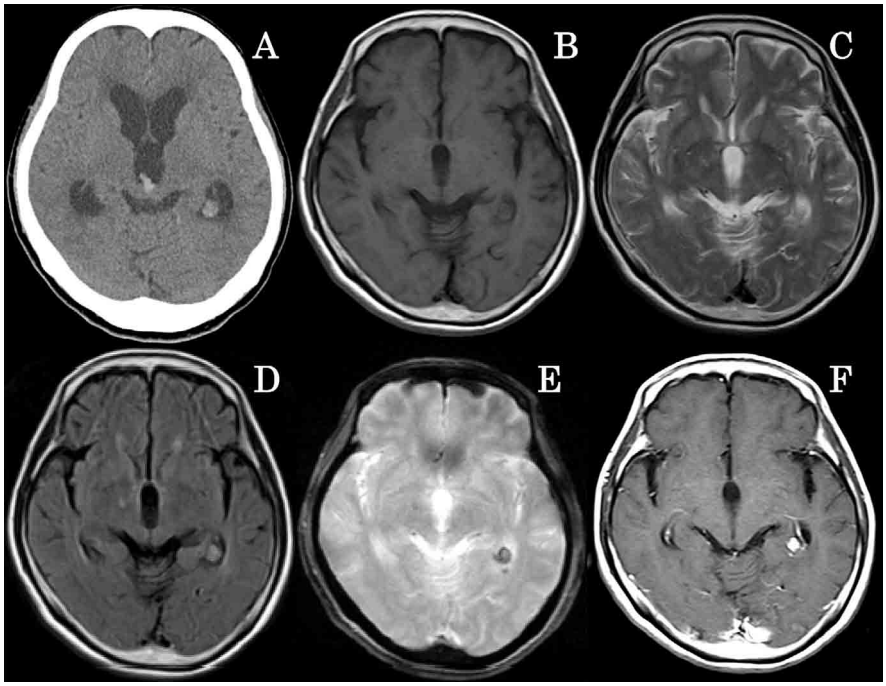

Figure 1 Admission CT scan showing a hyperdense lesion in the trigone of the left lateral ventricle, and a small mount of hemorrhage in the cerebral aqueduct along with acute hydrocephalus (A). The lesion was isointense on T1-weighted (B) ; hypointense on T2-weighted (C) ; hyperintense on FLAIR (D) ; 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).

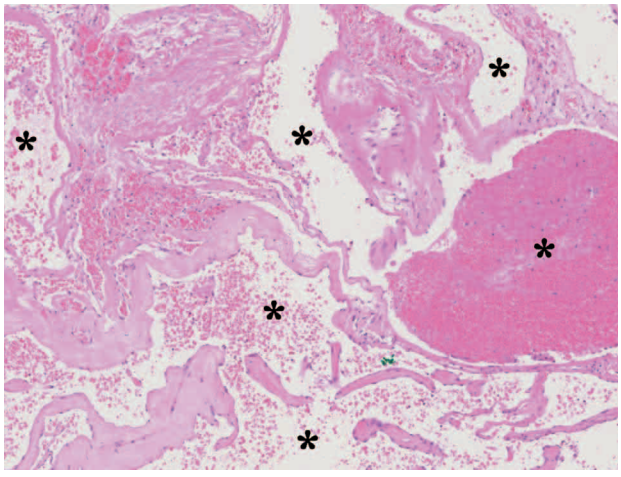

Figure 2 Dilated vessels (*) lined by endothelial cells with fibrous stroma. There was no glial tissue between the vessels. H & E, × 100

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 *et al.*	4	36	F	Seizures	Rt	NA	normal	hyperdensity	NA	Hemianopia
2	1977	Numaguchi *et al.*	22	43	M	Mass lesion	Rt	NA	venous stain and venous pooling	NA	NA	Hemiplegia, hemianopia
3	1979	Pau and Orynesu *et al.*	23	56	F	IVH	Lt	NA	not recognizable	NA	inoperable	Dead
4	1983	Iwasa *et al.*	24	8	F	Mass lesion	Lt	3	NA	hyperdensity	temporal craniotomy	Improvement
5	1983	Handa *et al.*	7	73	M	Mass lesion	Rt	3	no tumor stain	NA	lower posterior parietal lobe incision	Improvement
6	1986	Yamasaki *et al.*	25	73	M	Mass lesion	Rt	3	no feeding artery, tumor stain, draining vein	hyperdensity, calcification	NA	Improvement
7	1989	Andoh *et al.*	16	62	F	Mass lesion	Lt	3	no revealed homogenous overshadowing	hyperdensity	middle temporal gyrus	Dead
8	1990	Tatagiba *et al.*	7	35	M	Seizures	Rt	4	normal	hyperdensity	calcarine fissure	Improvement
9				24	M	Mass lesion	Lt	3	draining vein	hyperdensity	transcorticotomy (behind angular gyrus)	Improvement
10	1993	Miyagi *et al.*	17	3	F	Mass lesion	Lt	NA	no abnormal staining	hyperdensity	transsylvian transventricular	Improvement
11	2003	Nieto *et al.*	26	11	F	Seizures	Lt	5	NA	NA	temporal craniotomy	Improvement
12	2006	Kumar *et al.*	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	middle temporal gyrus	Hemiparesis
14				20	M	Mass lesion	Rt	5	no abnormality	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 *et al.*	18	52	F	Mass lesion	Rt	3	unremarkable	NA	low postcentral transcortical	Improvement
17	2012	Ohbuchi *et al.*		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 cereberal 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 hyposignal 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

REFERENCES

1. Simard JM, Garcia-Bengochea F, Ballinger WE Jr, Mickle JP, Quisling RG. Cavernous angioma : a review of 126 collected and 12 new clinical cases. Neurosurgery 18 : 162-172, 1986
2. Voci A, Panzarasa G, Formaggio G, Arrigoni M, Geuna E. Rare localizations of cavernomas : 4 personal cases. Neurochirurgie 35 : 99-101, 1989 (in French)
3. Reyns N, Assaker R, Louis E, Lejeune JP. Intraventricular cavernomas : three cases and review of the literature. Neurosurgery 44 : 648-655, 1999
4. Coin CG, Coin JW, Glover MB. Vascular tumors of the choroid plexus diagnosis by computed tomography. J Comput Assist Tomogr 1 : 146-148, 1997
5. Gerlach J. Intracerebral hemorrhage caused by microangioma. Progr Neurol Surg 3 : 363-396, 1969
6. Katayama Y, Tsubokawa T, Maeda T, Yamamoto T. Surgical management of cavernous malformations of third ventricle. J Neurosurg 80 : 64-72, 1994
7. Tatagiba Mes, Schonmayr R, Samii M. Intraventricular cavernous angioma. A survey. Acta Neurochir (Wien) 110 : 140-145, 1991
8. Kivelev J, Niemela M, Kivisaari R, Hernesniemi J. Intraventricular cerebral cavernomas : a series of 12 patients and review of the literature. J Neurosurg 112 : 140-149, 2010
9. Towfighi J, Bilaniuk LT, Zimmerman RA, Langfitt TW, Gonatas NK. Hemorrhages in bilateral choroid plexus hemangiomas demonstrated by computed tomography. J Neurosurg 45 : 218-222, 1976
10. Sigal R, Halimi P, Doyon D, Blas C, Chan KY. Imaging of cavernomas of the brain : Tomodensitometry and magnetic resonance imaing Neurochirurgie 35 : 89-94, 1989 (in French)
11. Kamrin RB, Buchsbaum HW. Largevascular malformation of the brain not visualized by serial angiography. Arch Neurol 13 : 413-420, 1965
12. Kawai K, Fukui M, Tanaka A, Kuramoto S, Kitamura K. Extracerebral cavernous hemangioma of the middle fossa. Surg Neurol 9 : 19-25, 1978
13. Pozzati E, Gaist G, Poppi M, Morrone B, Padovani R. Microsurgical removal of paraventricular cavernous angiomas : Report of two cases. J Neurosurg 55 : 308-311, 1981
14. Voigt K, Yasargil MG. Cerebral cavernous haemangiomas or cavernomas. Neurochirurgia (Stuttg) 19 : 59-68, 1976
15. Kumar GS, Poonnoose SI, Chacko AG, Rajshekhar V. Trigonal cavernous angiomas :

report of three cases and review of literature. Surg Neurol 65 : 367-371, 2006
16. Andoh T, Shinoda J, Miwa Y, Hirata T, Sakai N, Yamada H, Shimokawa K : Tumors at trigone of the lateral ventricle-Clinical analysis of eight cases. Neurol Med Chir (Tokyo) 30 : 676-684, 1990
17. Miyagi Y, Mannoji H, Akaboshi K, Morioka T, Fukuo M. Intraventricular cavernous malformation associated with medullary venous malformation. Neurosurgery 32 : 461-464, 1993
18. Stavrinou LC, Stranjalis G, Flaskas T, Sakas DE. Trigonal cavernous angioma : a short illustrated review. Acta Neurochir 151 : 1517-1520, 2009
19. Delandsheer JM. Meningiomas of the lateral ventricle. Neurochirurgie 11 : 3-83, 1965 (in French)
20. Delatorre E, Alexander E Jr, Davis CH Jr, Crandell DL. Tumors of the lateeral ventricles of the brain. Report of eight cases, with suggestions for clinical management. J Neurosurg 20 : 461-470, 1963
21. Handa H, Nagasawa S. Surgery of trigonal tumor. No shinkei Geka 12 : 901-912, 1984 (in Japanese)
22. Numaguchi Y, Fukui M, Miyake E, Kishikawa T, Ikeda J, Matsuura K, Tomonaga M, Kitamura K. Angiographic manifestations of intracereberal cavernous hemangioma. Neuroradiology 14 : 113-116, 1997
23. Pau A, Orunesu G. Vascular malformations of the brain in achondroplasia. Case report. Acta Neurochir (Wien) 50 : 289-292, 1979
24. Iwasa H, Indei I, Sato F. Intraventricular cavernous hemangioma. Case report. J Neurosurg 59 : 153-157, 1983
25. Yamasaki T, Handa H, Yamashita J, Paine JT, Tashiro Y, Uno A, Ishikawa M, Asato R. Intracranial and orbital cavernous angiomas. A review of 30 cases. J Neurosurg 64 : 197-208, 1986
26. Nieto J, Hinojosa J, Munoz MJ, Esparza J, Ricoy R. Intraventricular cavernoma in pediatric age. Childs Nerv Syst 19 : 60-62, 2003
27. Gonzalez-Darder JM, Pseudo-Martinez JV, Merino-Pena J. Trigonal cavernous angioma : case report. Neurocirugia (Astur) 18 : 330-332, 2007 (in Spanish)