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 PubMed 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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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*
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).

**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).
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 hypointensity 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
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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
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