**Fetal Cavernous Angioma**

—Case Report—

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**Abstract**

A neonate male who developed cavernous angioma was born with a severe intraventricular hemorrhage and intraparenchymal hemorrhage in the basal ganglia. Magnetic resonance imaging revealed a large, non-enhanced hematoma that packed the lateral ventricle. Surgery on the 2nd day of life verified a cavernous angioma associated with minimal hemosiderin and gliosis. Fetal cavernous angiomas, unlike such malformations in other age groups, can present with a devastating hemorrhage because of the lack of gliosis in the surrounding brain.

Key words: cavernous malformation, fetus, magnetic resonance imaging, hemorrhage

**Introduction**

Cavernous angioma is unusual in early childhood, and extremely rare in the neonatal period. We present a neonatal case with a severe intraventricular hemorrhage secondary to a basal ganglia cavernous angioma.

**Case Report**

A 29-year-old pregnant female was admitted to our hospital on July 7, 1994, because serial ultrasoundography after 32 weeks of gestation had revealed progressive hydrocephalus of the fetus. On July 10, 1994, a male infant at 36 weeks of gestation was delivered by cesarean section. The patient was admitted to the Newborn Intensive Care Unit of Nara Medical University. The family history was unremarkable.

Neurological examination revealed poor reactivity, macrocephaly, and a full, tense fontanelle. Ultrasonography through the anterior fontanelle revealed a large hydrogenic mass in the lateral ventricle that extended to the right caudate head (Fig. 1). On the 2nd day of life, hydrocephalus had progressed. Magnetic resonance (MR) imaging showed an intraventricular homogeneous mass occupying the right anterior horn and right caudate head, and extending to the posterior horn (Fig. 2). There was no enhancement by gadolinium (Fig. 3).

The patient underwent surgical removal of the mass and ventricular drainage on the 2nd day of life. The mass was approached via the right anterior horn after a right frontolateral craniotomy. An old, dark-red hematoma occupied the ventricle. The mass was totally removed. Histological examination of the specimen demonstrated a cavernous angioma (Fig. 4). The malformation was composed of thin-walled, dilated vascular spaces filled by thrombi at the same stage of organization and perilesional tissue that contained little hemosiderin and minimal gliosis.

Postoperatively, the patient experienced hydrocephalus and a generalized tonic seizure. Two months following surgery, the patient underwent a ventriculoperitoneal shunt. Computed tomography of the head 4 months after the first operation demonstrated ventricular dilation and right frontal lobe atrophy. The patient now has moderate mental retardation.

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Fig. 1 upper: Ultrasonogram, coronal view, on the 1st day of life demonstrating an intraventricular mass lesion (arrow) attached to the right caudate head. lower: Ultrasonogram, sagittal view, revealing an intraventricular mass (arrow) that extends to the posterior horn and marked hydrocephalus.

Fig. 2 T1-weighted magnetic resonance images, adjacent anterior (left) and posterior (right) coronal sections, on the 2nd day of life could not distinguish this lesion from a simple hematoma.

Fig. 3 T1-weighted magnetic resonance images, sagittal views, on the 2nd day of life showing the mass has two components, a high-intensity area (arrow) and another area (arrowheads) surrounding the high-intensity lesion and extending to the posterior horn (left). There was no gadolinium enhancement, and debris of the mass is lying in the bottom of the lateral ventricle (arrow) (right).

Fig. 4 Photomicrograph of the surgical specimen showing honeycomb-like, thin-walled vascular channels with no intervening neuroglial parenchyma. HE stain, ×100.

Discussion

The clinical data for the seven reported cases of neonatal cavernous angioma are summarized in Table 1.1,3,8,9,14

Neonatal cavernous angioma do not have a specific anatomic localization, but usually manifest as severe intracranial hemorrhage and have a poor prognosis.16 Histological examination of the perilesional parenchyma demonstrated minimal hemosiderin and gliosis in our case as compared to adult cases.
Gliosis in the surrounding brain in adult patients may exert a limiting membrane-like effect on a potential hemorrhage. In our case, the lack of such an effect seemed to contribute to the severe hemorrhage. We speculated that the large hemorrhages associated with fetal cavernous angiomas are due to the weakness of perilesional tissue.

Adult cavernous malformation contain more hemosiderin and hyalinized vessels. MR imaging shows a honeycomb appearance that is considered to be pathognomonic. However, the cavernous angioma in the present case was very difficult to distinguish from a simple hematoma on MR imaging. Cavernous angioma should be included in the differential diagnosis of severe intraparenchymal hemorrhage in neonates, even if the radiological findings suggest a simple hematoma. The clinical presentation and histological findings of neonatal cavernous angiomas are significantly distinct from adult malformations. Hemorrhages in the neonatal period can be particularly severe because of immature gliosis surrounding the angioma, and may present as neurological emergencies.

**References**

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