www.jkns.or.kr 10.3340/jkns.2009.45.2.99
Print ISSN 2005-3711 On-line ISSN 1598-7876
J Korean Neurosurg Soc 45 : 99-102, 2009

Case Report

Inflammatory Pseudotumor in the Lateral Ventricle with Repeated Bleeding-Case Report-

Jong-Hwa Park, M.D., Taek-Kyun Nam, M.D., Sung-Nam Hwang, M.D., Seung-Won Park, M.D.
Department of Neurosurgery, Chung-Ang University Yong-San Hospital, Seoul, Korea

Inflammatory pseudotumor is an uncommon lesion with unknown etiology characterized by sclerosing inflammation which clinically and radiographically mimics a neoplastic lesion. A 47-year-old man presented with sudden headache and dysarthria. Brain CT scan revealed a 2.6 × 2.2 cm sized, round, and hyperdense mass in the anterolateral wall of the left lateral ventricular trigone. On MR imaging studies, the mass showed low signal intensity in the wall of the trigone on T2-weighted image, central mixed (iso- and high-) signal intensity with peripheral low-signal intensity on T1-weighted image. Subtle staining of left choroid plexus with irregular shaped distal branch of anterior choroidal artery was found on the cerebral angiography. These findings suggested a small tumorous lesion originated from the left choroid plexus. During the hospital days, the mass manifested as repeated hemorrhages. The mass was successfully removed via left occipital transcortical approach. The histopathological report of the specimen was hemorrhage and fibrosis, with dense lymphoplasma cell infiltration, suggestive of an inflammatory pseudotumor.

KEY WORDS : Intraventricular tumor · Inflammatory pseudotumor · Hemorrhage.

INTRODUCTION

Inflammatory pseudotumor is an uncommon lesion with unknown etiology characterized by sclerosing inflammation which clinically and radiographically mimics a neoplastic lesion[2,4,7]. Although CT and MR imaging characteristics may simulate a neoplastic mass, pathologically it is composed of inflammatory cells.

We present a rare case of a patient with an inflammatory pseudotumor which bled repeatedly and review the literatures regarding clinical, radiological, and pathologic aspects.

CASE REPORT

A 47-year-old man presented with headache and dysarthria which developed 4 days and aggravated 1 day before admission. He did not have any remarkable past medical problems. Initial brain CT scan demonstrated a 2.6×2.2 cm sized, round hyperdense mass like lesion in the anterolateral wall of trigone of the left lateral ventricle, which was not enhanced prominently (Fig. 1A, B). On MR imaging studies, the mass showed low signal intensity in the wall of the trigone on T2-weighted image, central mixed (iso- and high-) signal intensity with peripheral low-signal intensity on T1-weighted image (Fig. 2). It was indistinct whether the mass involved the choroid plexus in the left trigone or not. Subacute hematoma adjacent to the mass was noted in the left periventricular white matter. Considering these CT, MR findings and clinical history of aggravated headache and dysarthria, an acute hemorrhage within the mass lesion with adjacent subacute hematoma was suspected.

On 12th day after admission, the patient complained of aggravated headache in spite of the conservative management. Follow-up brain CT was checked and revealed a slight enlargement of the mass due to rebleeding. On 25th day after admission, the dysarthria was worsened into sensorimotor aphasia and finally his consciousness was deteriorated into stupor. On follow-up brain CT, the mass size increased more by repeated bleedings and peripheral swelling around the mass increased (Fig. 1C, D).

Preoperative cerebral angiography showed subtle hyperdense staining of left choroid plexus with irregular shaped

• Received : June 16, 2008 • Accepted : January 27, 2009
• Address for reprints : Taek-Kyun Nam, M.D.
Department of Neurosurgery, Chung-Ang University Yong-San Hospital, Hangang-no 3-ga, Yongsan-gu, Seoul 140-757, Korea
Tel : +82-2-748-9717, Fax : +82-2-798-2818
E-mail : nsnam@dreamwiz.com

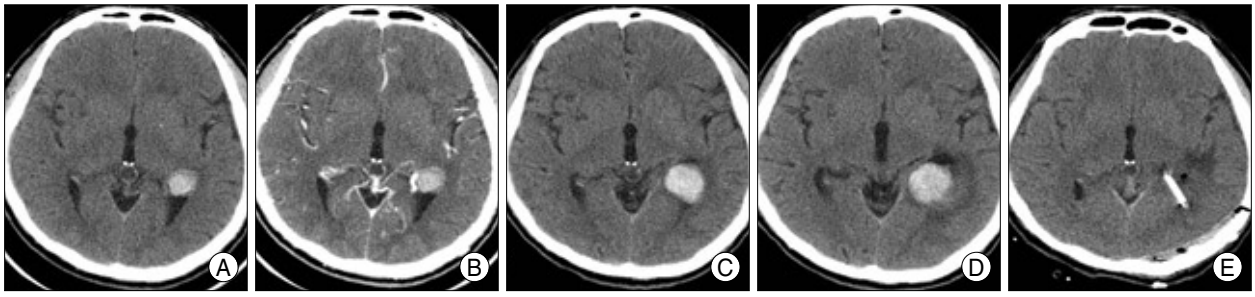

Fig. 1. A and B : Brain computed tomography (CT) scan at admission shows a hyperdense mass like lesion in the anterolateral wall of trigone of the left lateral ventricle. This lesion was not enhanced prominently. C and D : Follow-up CT scans show repeated bleeding and enlarged mass. E : Postoperative CT scan shows the complete removal of the mass and hematoma.

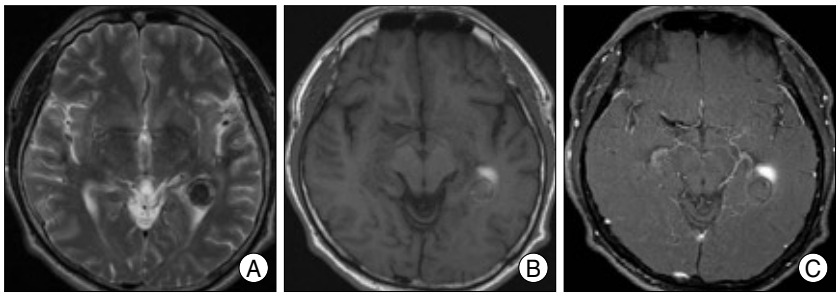

Fig. 2. A : Brain magnetic resonance (MR) image shows a 2.6 x 2.2 cm sized round mass in the wall of the left trigone with dark signal intensity in T2-weighted image. B : The mass reveals central mixed (iso- and high-) signal intensity with peripheral low-signal intensity on T1-weighted image, and 1.2 cm sized subacute hematoma adjacent to the mass is noted in the left peritrigonal white matter. C : Gadolinium-enhanced T1-weighted MR image shows barely enhanced mass lesion.

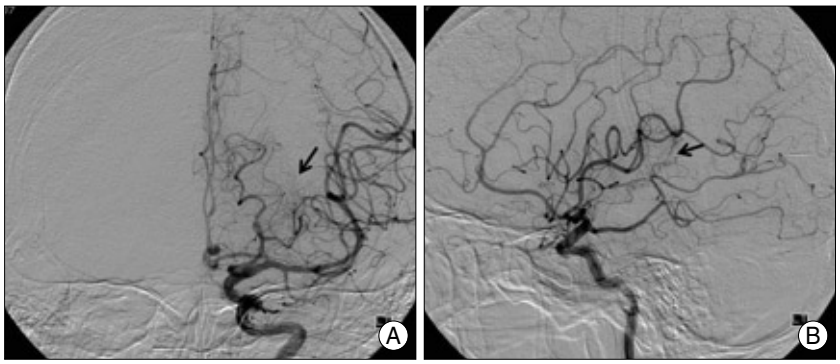

Fig. 3. Cerebral angiography. Subtle hyperdense staining of left choroid plexus with irregular shaped distal branch of anterior choroidal artery (arrow) was found. A : AP view. B : Lateral view.

distal branch of anterior choroidal artery (Fig 3). These findings suggested a small tumorous lesion in the left choroid plexus.

The mass was successfully removed via left occipital transcortical approach (Fig. 1E). The hematoma was located in the left lateral ventricle. After removal of the hematoma, a brownish mass was found adhered to the ventricular wall and choroid plexus. It was dissected from the ventricular wall, and there were several arterial feeders from the choroid plexus. The main arterial feeder was at the anteroventral portion of the mass. The mass was excised en bloc, which size was measured 2 cm × 1.5 cm × 1.4 cm.

The histopathological examination of the specimen revealed a hematoma and fibrosis, with dense lymphoplasma cell infiltration, which findings were corresponding with an inflammatory pseudotumor (Fig. 4). No mitosis, necrosis, or nuclear pleomorphism was noted.

The patient recovered to be alert and the headache and sensorimotor aphasia were gradually improved after the operation. He was discharged with minimal sensorimotor dysarthria.

DISCUSSION

Inflammatory pseudotumor is a pathologic diagnosis which consists of a reactive, inflammatory, non-neoplastic proliferation of connective tissue. It is clinically and radiographically indistinguishable from invasive neoplasms. Differential diagnosis should include an inflammatory pseudotumor when there is a localized, solitary, nontumorous, and space-occupying lesion. Inflammatory pseudotumor is comprised of intermingling collagen fibers, inflammatory cells such as lymphocytes and/or plasma cells, and mesenchymal cells such as fibroblasts and myofibroblasts[6]. It destroys the structure from which it is originated[6].

The pathophysiology of inflammatory pseudotumor remains obscure. It is said to be caused by an exaggerated immunological process and sometimes associated with high level of serum immunoglobulin[1]. But, the role of chronic infection or immunologic response has not been clearly

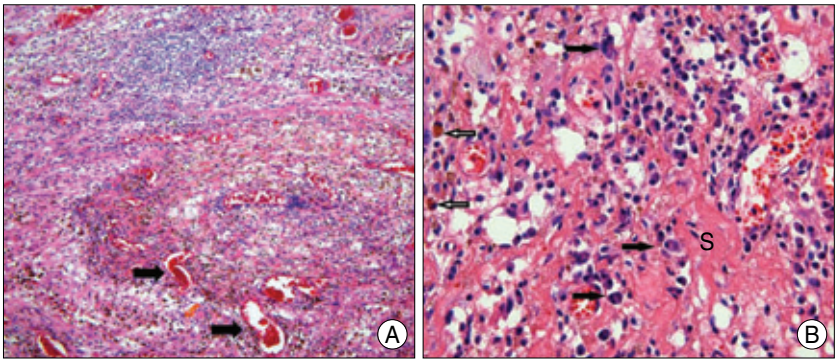

Fig. 4. Pathological findings. A : The lesion shows marked vascular proliferation (black arrow). (×100, H&E stain). B : Dense lymphoplasma cell infiltration (black arrow), mixed with hemosiderin-laden macrophages (white arrow) in the fibrotic stroma (S) (×400, H&E stain).

Table 1. Comparison of previous reported cases of inflammatory pseudotumor of the choroid plexus

Characteristic	Previous 5 cases*	Current case
Location	Lateral ventricle (4), 4th ventricle (1)	Lateral ventricle
Symptoms	Neurologic deterioration and hemiparesis (4)	Headache, dysarthria
	Headache, diplopia, vomiting (1)	
Neuroimaging findings	Contrast enhancement : prominent	Contrast enhancement : not prominent
	Hemorrhage : none	Hemorrhage : repeated bleeding
Treatment	Excision (4), open biopsy (1)	Excision
Outcome	Good recovery (2), Moderate disability (1)	Good recovery
	Died of pulmonary embolism (1)	
	Died of gastric ulcer (1)	

*Chang et al. 1991, Pimental et al. 1993, al-Sarraj et al. 1995, Bramwit et al. 1997, and Miyahara et al. 2008

elucidated. Bacterial or viral infection has also been postulated as a cause of a orbital pseudotumor[8].

Inflammatory pseudotumor has been described most often in the lung, but less often in other organs. Primary central nervous system (CNS) involvement is relatively rare[5,7]. Inflammatory pseudotumor of the CNS is a condition often affecting young adults with a slight male preponderance[7]. Miyahara et al.[6] reported that there have been only 5 case reports of inflammatory pseudotumor that developed in the choroid plexus (Table 1). In these cases, alterations in consciousness and contralateral hemiparesis were the initial symptoms, and the masses showed a contrast enhancement. In our patient, headache and dysarthria were the first manifestations, and there were repeated hemorrhages in the mass. Our case might be the first case of an inflammatory pseudotumor with repeated hemorrhages in the lateral ventricle (Table 1).

There are many differential diagnosis of a well-circumscribed enhancing intraventricular mass in the lateral ventricle, such as meningioma, metastasis, xanthogranuloma, neurosarcoidosis, oligodendroglioma, astrocytoma, ependymoma, choroid plexus papilloma, and neurocytoma[2]. In differentiating these from inflammatory pseudotumor, the patient's age, the location and imaging characteristics of the lesion will be helpful. Choroid plexus papilloma in the lateral ventricle is more common in younger patients. Ependymomas are more common in the 4th ventricle. Metastasis is strongly suspected when there is a history of primary neoplasm. Intraventricular neurocytoma and oligodendroglioma are sometimes difficult to distinguish, but neurocytoma is frequently attached to the septum pellucidum. Xanthogranuloma, neurosarcoidosis, and low-grade astrocytoma are unlikely to bleed[2]. In previously reported cases of intraventricular pseudotumor, there also was no tumor bleeding[1-3,6,8]. However, in our case, repeated bleedings was characteristic.

Although inflammatory pseudotumor has no distinctive characteristics either clinically or radiographically, we suggest that it should be included in the differential diagnosis of intraventricular mass.

CONCLUSION

We present a rare case of inflammatory pseudotumor of the lateral ventricle with repeated bleeding. Inflammatory pseudotumor should be considered in the differential diagnosis of intraventricular mass lesions.

References

1. Al-Sarraj S, Wasserberg J, Bartlett R, Bridges LR : Inflammatory pseudotumour of the central nervous system : Clinicopathological study of one case and review of the literature. **Br J Neurosurg 9** : 57-66, 1995
2. Bramwit M, Kalina P, Rustia-Villa M : Inflammatory pseudotumor of the choroid plexus. **AJNR Am J Neuroradiol 18** : 1307-1309, 1997
3. Chang Y, Horoupian DS, Lane B, Fross RD, Smyth LT Jr , Seiling RJ : Inflammatory pseudotumor of the choroid plexus in sjogren's disease. **Neurosurgery 29** : 287-290, 1991
4. Chen JM, Moll C, Schotton JC, Fisch U : Inflammatory pseudotumors of the skull base. **Skull Base Surg 4** : 93-98, 1994
5. Mirra SS, Tindall SC, Check IJ, Brynes RK, Moore WW : Inflammatory meningeal masses of unexplained origin. An ultrastructural

and immunological study. **J Neuropathol Exp Neurol 42** : 453-468, 1983
6. Miyahara K, Fujitsu K, Yagishita S, Takemoto Y, Ichikawa T, Matsunaga S, et al : Inflammatory pseudotumor of the choroid plexus. **J Neurosurg 108** : 365-369, 2008
7. Olivares-Romero J, Serrano-Castro PJ, Roig JM, Guardado-Santervas P, Peralta-Labrador JI, Goberna-Ortiz E : [Inflammatory pseudotumor : differential diagnosis of tumors of the 4th ventricle.] **Rev Neurol 32** : 63-67, 2001
8. Pimentel J, Costa A, Tavora L : Inflammatory pseudotumor of the choroid plexus. **J Neurosurg 79** : 939-942, 1993