

## Meningioma *en Plaque* of Parasagittal Region Presented with Recurrent Venous Infarction

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A case of parasagittal meningioma *en plaque* with a peculiar clinical presentation is reported with a review of the literature. A 72-year-old woman presented with dysphasia and right hemiparesis. Computed tomography and magnetic resonance imaging demonstrated focal edema of left frontal lobe and a thick sheet-like parasagittal enhancing lesion with extension along the falx cerebri and adjacent sulcal enhancement. Differential diagnosis included idiopathic hypertrophic pachymeningitis, meningeal neurosarcoidosis, metastasis and meningioma *en plaque*. Cerebral angiography revealed occlusion of the anterior one-third of the superior sagittal sinus as well as a faint tumor blush supplied from the anterior branch of the middle meningeal artery. At surgery, the tumor invading the dura and skull was removed totally but the tumor invaded into the superior sagittal sinus was removed subtotally. The tumor was confirmed to be a transitional meningioma on pathological examination.

**KEY WORDS :** Meningioma *en plaque* · Superior sagittal sinus occlusion · Cerebral venous infarction · Cerebral angiography.

### Introduction

Meningioma *en plaque* (MEP) is a tumor of limited thickness that grows along the planes of the meninges and, in some cases, occupies a considerable area<sup>4,10</sup>. MEP usually occur at the middle cranial fossa, most commonly involving the sphenoid bone. It has rarely been reported in other parts of the skull<sup>4,5,8,10,11,18,20,22</sup>. To our knowledge, only one case of parasagittal meningioma *en plaque* has been reported<sup>1</sup>. We report a case of parasagittal meningioma in a 72-year-old woman with unusual clinical presentation of recurrent venous infarction with a review of the literature.

### Case Report

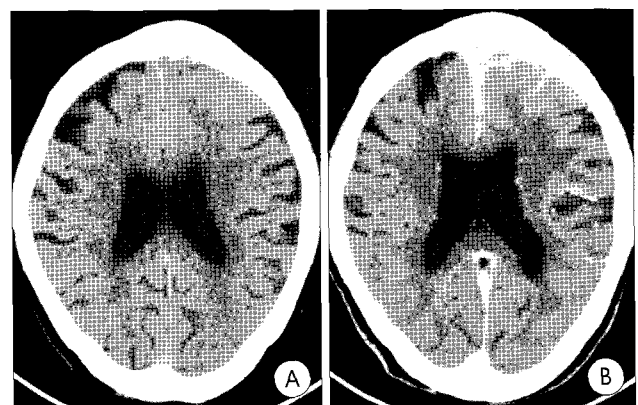
#### Clinical presentation

A 72-year-old woman was admitted for evaluation of right hemiparesis and dysphasia which had been developed three weeks ago and then was aggravated a day prior to admission. She had no previous history of medication, surgery or trauma. On neurologic examination, she showed alert mentality, motor

dysphasia and right hemiparesis of grade 4/5 without abnormality in sensation.

#### Hospital course & diagnostic evaluation

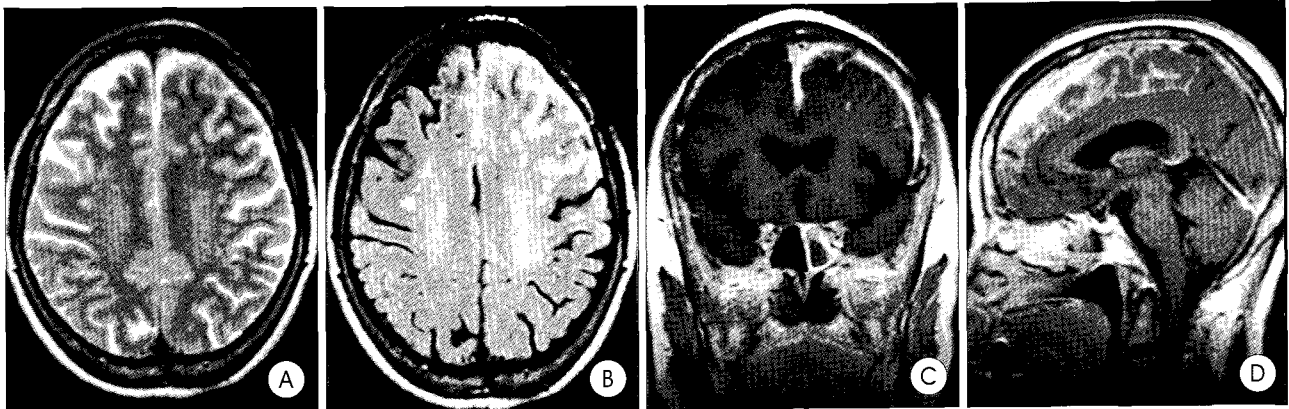
Computed tomography (CT) disclosed gyral swellings in the left frontal lobe and a linear enhancing lesion that extended



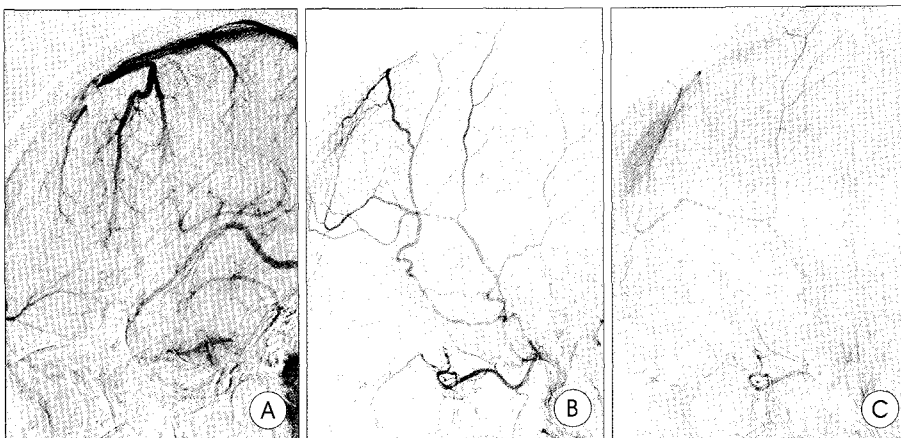
**Fig. 1.** Computed tomography (CT) shows gyral swellings in the left frontal lobe with localized hyperostosis (A) and enhanced CT shows a linear enhancing lesion that extended along the interhemispheric fissure, consistent with meningeal thickening (B).

• Received : June 5, 2006 • Accepted : August 22, 2006

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**Fig. 2.** On magnetic resonance imaging, an axial T2-weighted image shows multiple focal hyperintense lesions in the left frontal lobe and both centrum semiovale (A), an axial FLAIR image shows cortical and subcortical hyperintense lesions with gyral swelling in the left frontal lobe (B), an enhanced coronal T1-weighted image shows parasagittal mass in the left frontal lobe extending along the falx cerebri (C) and an enhanced sagittal T1 weighted image shows sulcal enhancement adjacent to the linear sheet-like mass (D).



**Fig. 3.** Cerebral angiography reveals occlusion of the anterior one-third of the superior sagittal sinus (A) as well as the persistent tumor staining (arrows) from the anterior branch of the middle meningeal artery in the early (B) and late (C) arterial phases.

along the interhemispheric fissure, consistent with meningeal thickening (Fig. 1). Focal hyperostosis was also noted over the left frontal bone. Magnetic resonance (MR) imaging demonstrated a thick sheet-like parasagittal mass in the left frontal lobe extending along the falx cerebri (Fig. 2). Sulcal enhancement was present adjacent to the mass. Multiple patchy hyperintense lesions were shown in the FLAIR sequence. Differential diagnosis included idiopathic hypertrophic pachymeningitis, meningeal neurosarcoïdosis, metastasis and MEP.

At lumbar puncture, the opening pressure was 13cmH<sub>2</sub>O and the cerebrospinal fluid looked clear. Laboratory study disclosed pleocytosis (123 leukocytes/mm<sup>3</sup>), which comprised of 69% monocytes, 28% macrophages, and 3% polymorphonuclear leukocytes. No tumor cell was found. Total protein was 45mg/dL and glucose was 59mg/dL. Culture studies and immunological tests were negative for bacteria, fungi and various viruses.

Remarkable symptomatic improvement was attained by steroid medication. However, the effect of steroid was only te-

mporary and her symptoms were aggravated over a few days. Follow-up MR imaging revealed enlargement of the high signal intense lesions. Under the impression of recurrent venous infarction, cerebral angiography was performed and revealed occlusion of the anterior one-third of the superior sagittal sinus as well as a faint tumor blush from the anterior branch of the middle meningeal artery (Fig. 3).

### Operative findings

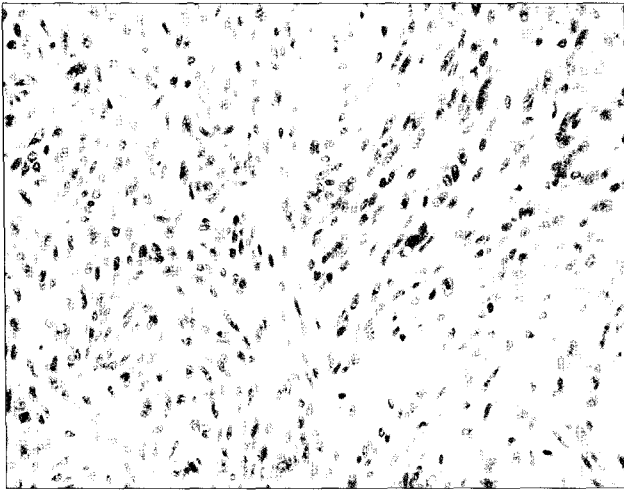
To establish the pathologic di-

agnosis she underwent operation. Under general anesthesia the left frontal craniotomy was done. Focal hyperostosis was noted on the outer surface of the skull, as was on CT. The inner surface of the skull was diffusely invaded by the tumor.

The dura was thickened, being more than 3.0mm in the thickest part. Moreover, the tumor invaded the falx cerebri and the superior sagittal sinus. The tan pinkish sheet-like mass was found adhered to the dura. The main mass including the thickened dura was removed and the invaded part of the superior sagittal sinus was partially removed and cauterized. The dural and skull defects were reconstructed with the artificial dura and with bone cement.

### Histological examination

Histologically, the tumor had lobular arrangements of fibrous and meningotheelial cells with mild nuclear atypia, which was diagnosed as a transitional meningioma according to the 2000 WHO classification of tumors of the nervous system<sup>13</sup> (Fig. 4). The tumor invaded the dura and the skull bone (Fig. 5).



**Fig. 4.** Histological features of meningioma shows transitional features of the fibrous and typical meningothelial cells with intranuclear inclusions. (H&E, x400).

#### Postoperative course

No further neurologic deficit was observed after operation and conventional radiation therapy (RT) was given. She was discharged home, and her condition gradually improved over several months after operation and RT.

#### Discussion

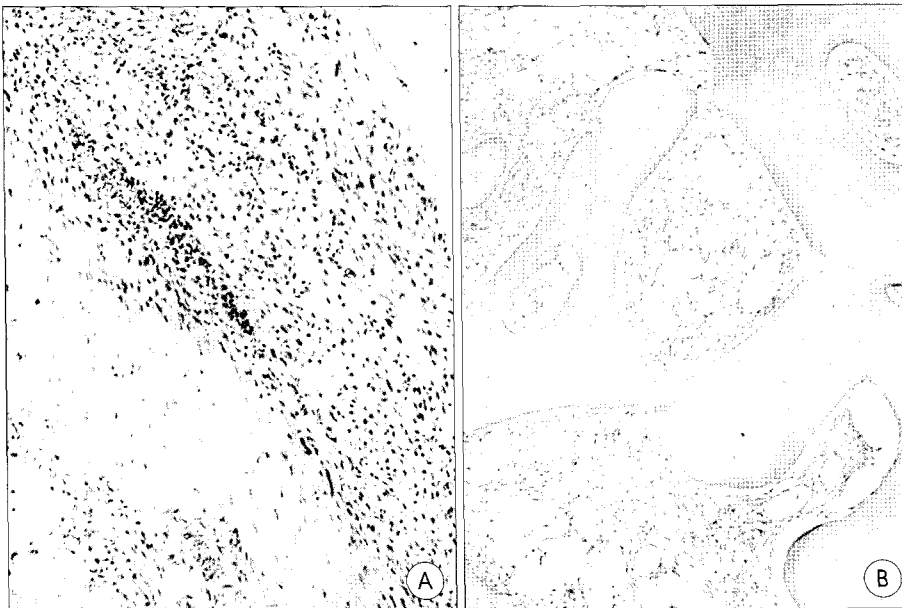
Meningioma *en plaque* is a rare tumor characterized more by its clinical and biological behavior than by its histological appearance<sup>3</sup>. MEP grows as a flattened appearance, not globoid as in meningioma *en masse* and spreads along the plane of meninges. It usually occurs at the skull base, especially along the sphenoid ridge and, rarely, at the convexity in the

vicinity of the coronal suture<sup>4,5,8,10,11,18,20,22</sup>. Parasagittal location, as was in our case, seems to be extremely rare<sup>1</sup>.

MEP almost occurs in women of middle age and frequently provokes bony hypertrophy that produces clinical signs and symptoms by compressing adjacent structures<sup>4,5,8,10,20</sup>. The duration of symptoms is usually protracted because of the minimal discomfort produced<sup>4,5,20</sup>. Proptosis, visual disturbance, facial or scalp swellings and headache are common clinical presentations. However, presentation with cerebral venous infarction in the cases of MEP has not been reported to our knowledge. In our case, there was occlusion of the superior sagittal sinus by the tumor invasion without adequate collateral drainage route, resulting in venous infarction. It could be differentiated from the peritumoral edema; clinically, the patient's symptoms were aggravated despite of steroid injection, and radiologically, cerebral angiography showed obstruction of the superior sagittal sinus, and MR imaging demonstrated hyperintense signal with gyral swelling and venous engorgement.

The differential diagnosis for a lesion causing dural thickening should include osteoblastic metastasis, idiopathic hypertrophic cranial pachymeningitis, and sarcoid granuloma, as well as MEP<sup>3,12,14,15,19,21,23</sup>. Fungal infection, such as coccidioidomycosis and blastomycosis, and tuberculoma also should be included in the differential diagnosis for a lesion of dural thickening and pleocytosis in the cerebrospinal fluid<sup>2,7,14</sup>. In the presence of hyperostosis, although it may be a clue to the diagnosis of meningioma, other hyperostosing conditions such as fibrous dysplasia or osteoma should be considered in the differential diagnosis<sup>10</sup>.

Although the exact mechanism of hyperostosis in meningioma remains unclear, tumor invasion of the bone is generally accepted among many hypotheses<sup>1</sup>. Kim et al.<sup>10</sup> suggested that the hyperostotic patterns associated with MEP were similar to those of globoid meningiomas. However, hyperostosis is frequently observed in MEP with an incidence of 13% to 49% while the incidence of hyperostosis is reported to be 4.5% of all the types of meningioma<sup>9</sup>. Hyperostosis in MEP was classified as four different patterns as follows: 1) homogenous patterns, 2) periosteal pattern, 3) three-layer pattern and 4) diploic pattern<sup>10</sup>. In our case, CT demonstrated three-layer pattern of hyperostosis, surface irregularity of the hyperostotic bone and adjacent intracranial c-



**Fig. 5.** The tumor invading the dura (A) and the skull vault (B) is demonstrated. (H&E, x400).

rescent-shaped enhancing lesion, but no inward bulging of the lesion was shown.

In a series of pterional MEP, the pathological type was psammomatous meningioma in the majority, while the meningothelial type was dominant in another review of convexity meningioma en plaque<sup>4,17</sup>. Our case was confirmed to be the transitional type. There was no relationship between the hyperostotic pattern and histologic type of MEP in a report<sup>10</sup>.

Previously, it was believed that patients with MEP should not be operated and surgical management should be considered only as a last resort because of slow progression, technical difficulty and high surgical mortality rate<sup>4,5,8</sup>. Nowadays, a fronto-temporal approach with lateral cranial base resection is advocated as treatment of choice in skull base MEP<sup>6,20</sup>. A hyperostosing MEP in the cranial vault is readily amenable to radical resection<sup>10</sup>. It is emphasized that the dura should be incised, the subdural space be explored even if it looks normal, and all portions involved by the tumor be removed<sup>5,10</sup>.

Although the role of radiation therapy in MEP has yet to be established, a number of reports recommended postoperative RT for cases of incomplete tumor resection and for cases of high-grade meningiomas<sup>5,8,10,16,20,22</sup>. According to Maroon et al.<sup>16</sup>, there was no tumor progression in 10 patients who received RT with a follow-up period ranging from 16 to 95 months. In our case, the patient received postoperative RT and recurrence has not been noted for 11 months.

## Conclusion

We present a rare case of parasagittal MEP with a review of the literature. Meningioma *en plaque* should be included in differential diagnoses in cases showing parasagittal dural thickening and recurrent venous infarction.

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