CASE REPORT

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Convexity Meningioma En Plaque Presenting with Diffuse Hyperostosis of the Skull

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Although meningioma is a common and benign intracranial tumor, meningioma en plaque is a rare tumor, especially in the cranial vault. Meningioma en plaque(MEP) usually occurs in the area of the sphenoid wing, and it causes cosmetic and visual problems, as well as the problems that are due to its mass effect. The authors present here a case of convexity meningioma en plaque that involved the skull and scalp with diffuse hyperostosis as the presenting salient radiological findings, which caused marked intraoperative bleeding.

KEY WORDS : Meningioma en plaque · Hyperostosis · Intraoperative bleeding.

Introduction

About a half of all the intracranial meningiomas locate in the skull base region[6]. Meningioma en plaque usually develop in the skull base and these are rare tumors that are characterized more by their clinical and biological behaviour than by their histological appearance[4]. Meningioma en plaque with extracranial extension can often be seen in the skull base, especially at the sphenoid wing[2-4,7]. We report here on a case of convexity meningioma en plaque without skull base involvement. This tumor had invaded the skull and scalp with diffuse hyperostosis that presented as the striking raiological findings, and this finding can be suggestive for marked intraoperative bleeding.

Case Report

Clinical history

A 44 year-old woman presented with headache, left hemiparesis (Grade IV-), and left temporal homonymous hemianopsia on september, 2003. Her past medical history was nonspecific. The neurological examination revealed mild left facial palsy and left hemiparesis. Left temporal homonymous hemianopsia was also found.

Preoperative radiological findings

A anterior-posterior skull X-ray film demonstrated bizarre, diffuse, significant hyperostosis and sclerosis in the cranial vault (Fig. 1A), and a lateral skull X-ray film showed erosion of the sella turcica (Fig. 1B). The magnetic resonance(MR) T2-weighted imaging revealed significant thickening of the skull and scalp, and there were signal voids in the scalp. The

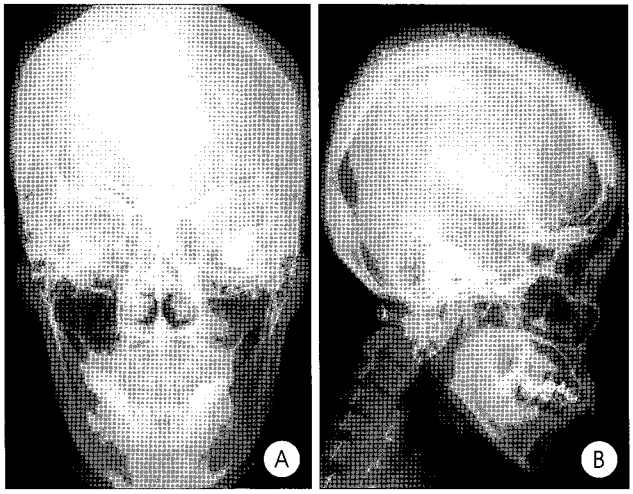

Fig. 1. The antero-posterior skull X-ray film demonstrates bizarre, diffuse, significant hyperostosis and sclerosis in the cranial vault (A), and the lateral skull X-ray film shows pressure atrophy (erosion) of the sella turcica (B).

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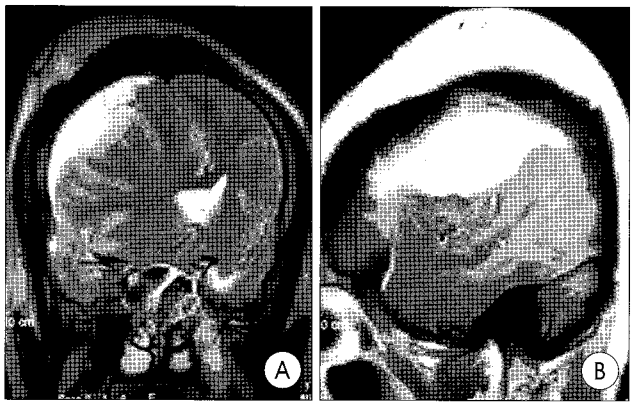

Fig. 2. The magnetic resonance (T2WI-coronal (A), T1WI-sagittal, enhanced (B)) imaging revealing significant thickening of the skull bone and the scalp and a carpet-like frontoparietal, well-enhanced heterogeneous mass. Signal voids in the scalp are noted. The tumor extends diffusely beneath the inner table.

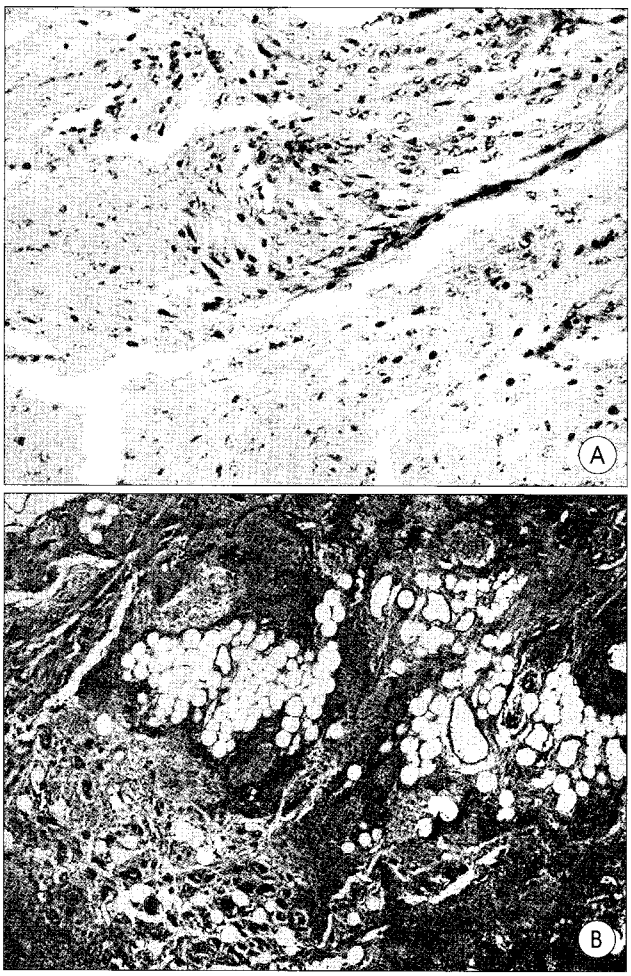

Fig. 3. Light microscopic findings reveal transitional meningioma without anaplasia (A) (H&E ×100). Tumor invasion to the subcutaneous adipose tissues and vessels of the scalp is present (B) (H&E ×40).

tumor tended to form a layer that closely followed the contour of the inner table of the skull for a considerable area. On the T1 weighted gadolinium enhanced image, the tumor was well enhanced in the frontoparietal region, and the tumor was the thickest in the right parasagittal region (Fig. 2). Those findings were compatible with meningioma en plaque. Cerebral angiography showed that the tumor was fed by the right middle meningeal artery, the superficial temporal artery, the occipital artery and it was also fed by the left middle meningeal artery and the superficial temporal artery. On October 2003, preoperative embolization of the occipital artery was done, which was one of the feeder arteries for the tumor.

Operative findings

As shown on the preoperative MRI, a dilated, and twisted scalp vessel and the hyperostotic woven bone were noted at the site of the meningioma. The scalp incision and craniotomy were performed which caused excessive blood loss that was unlike as the usual operation for convexity meningioma. The tumor, including the hyperostotic skull, was resected. Tight adhesion between the thickened scalp and the hyperostotic skull was noted. As soon as the scalp flap was separated from the underlying hyperostotic bone, marked bleeding was developed from the scalp and from the diffuse hyperostotic bone. The tumor had invaded not only the hyperostotic skull but also the scalp. The skull was fragile like woven bone because of diffuse invasion of the tumor, but it was not adhered to the dura mater at the operative site. The dura mater was hypervascular. Severe adhesion was present between the dura and the underlying tumor at the convexity, suggesting that the tumor had originated from the dura. The tumor was partially adhered to the underlying cortex. Most of the dura including the tumor was removed. But the tumor and dura adjacent to the superior sagittal sinus was electrocoagulated instead of being resected. The diffuse hyperostotic skull was also removed and cranioplasty was done. Intraoperatively, the patient received transfusions of 8 units of packed red cell(PRC) and 3 units fresh frozen plasma(FFP).

Histological examination

Microscopically, the tumor was composed of cells with delicate, round or oval nuclei, inconspicuous nucleoli, lightly eosinophilic cytoplasm and indistinct cytoplasmic borders. The tumor infiltrated into osseous trabeculae, and scalp making them thickening. The histological examination revealed transitional meningioma without anaplasia (Fig. 3).

Postoperative course

At the immediate postoperative period, the patient's neurological condition was unchanged compared to preoperative status, except for the slight deterioration of left hemiparesis (Grade III). She was discharged home on postoperative Day 14 with the same to preoperative left hemiparesis (Grade IV$^-$).

The patient's left hemiparesis was improved (Grade IV$^+$) at 3 months after the operation.

Discussion

Meningiomas en plaque(MEP) has been well described in the literature and it is often associated with hyperostosis of the skull[1]. MEP is commonly located in the middle cranial fossa and the sphenoid wing, and it is rarely found at other sites[3,7]. The tumor is typically slowgrowing, and it has a flat or slightly nodular shape for a considerable area. Hyperostosis is frequently observed in MEP with an incidence of 13% to 49%, whereas this finding makes up 4.5% of all the types of meningioma[5]. In the case of our patient, the tumor diffusely extended into the subdural space beneath the calvaria, and diffuse hyperostosis of the cranial vault was also present. Further, our case demonstrated that the tumor had infiltrated into the scalp. Although the exact mechanism of the hyperostosis in meningioma remains unclear, several theories have been proposed as follows; vascular disturbance of the bone that is caused by the tumor, irritation of the bone by the tumor without invasion, stimulation of the osteoblasts in the normal bone by the factors secreted by the tumor cells, production of bone by the tumor itself, and tumor invasion of the bone[8]. In our case, the tumor cells had diffusely invaded into the hyperostotic bone and scalp via the haversian canal, and this suggested that the diffuse hyperostosis was caused by the tumor invasion.

Conclusion

Although it is rare that MEP demonstrates diffuse massive hyperostosis, clinicians should be aware of this possibility. In author's experience, it is important to localize the midline structure such as superior sagittal sinus before the craniotomy due to distortion of normal anatomy by marked hyperostosis. And care should be attentively taken to prevent excessive blood loss during the operation, and particularly when removing the hyperostotic bone flap and when dealing with the tumor that has invaded the superior sagittal sinus.

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