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Case Report

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An Osteolytic Meningioma en Plaque of the Sphenoid Ridge

Meningioma en plaque (MEP) is a rare tumor characterized more by its clinical and biological behavior than its histological appearance. Hyperostosis of the skull is one of the characteristic signs of MEP. This bony change can produce clinical symptoms and signs in MEP by pressing against adjacent structures. The authors report a rare case of an osteolytic MEP extending from the sphenoid wing into the orbital wall, middle fossa, and temporalis muscle.

KEY WORDS : Meningioma en plaque · Osteolytic.

INTRODUCTION

The term, meningioma en plaque (MEP), was coined by Cushing and Eisenhardt to differentiate this type of tumor from the more common form, which is called meningioma en masse[7]. MEP is a rare tumor associated with hyperostosis of the skull. MEP represents a morphological subgroup defined by a carpet or sheet-like lesion that infiltrates the dura and sometimes invades the bone[8]. It is more likely to provoke adjacent bony hyperostosis, and the amount of hyperostosis is often disproportionate to the size of the relatively small tumor[6,10,14,19,25]. This bony change is a characteristic sign of MEP[19] and it produces the clinical symptoms and signs[3,6,9]. The authors report a rare case of an osteolytic, not hyperostosing MEP extending from the sphenoid wing into the orbital wall, middle fossa, and temporalis muscle.

CASE REPORT

A 74-year-old male complained of right proptosis, diplopia and headache that had persisted for 6 months. He had been treated for hypertension for several years. He had no previous history of trauma. On ophthalmologic examination, visual acuity and extraocular eye movement were normal. The patient had no neurological deficits. Magnetic resonance imaging (MRI) revealed a contrast-enhanced extra-axial mass in the right sphenoid ridge area. This mass showed intraorbital invasion through the lateral wall of the right orbit and soft tissue mass in the medial aspect of the right temporalis muscle (Fig. 1). Computed tomography (CT) revealed irregular osteolytic changes with some cortical disruption of the right sphenoid wing (Fig. 2).

The patient underwent a surgery using a modified orbito-zygomatic approach. The osteolytic lesion at the sphenoid bone was removed by using a high-speed drill and rongeur. The lateral wall of the orbit was removed, and the superior orbital fissure was decompressed. The bone

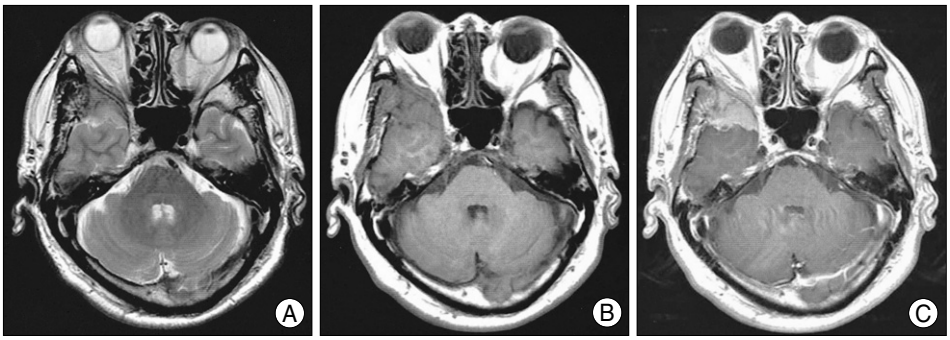

Fig. 1. Axial T2-weighted (A) and T1-weighted (B) images show an extraaxial mass in the right sphenoid ridge area. This mass extends lateral wall of right orbit and medial aspect of right temporalis muscle. After contrast (C), strong enhancement is seen.

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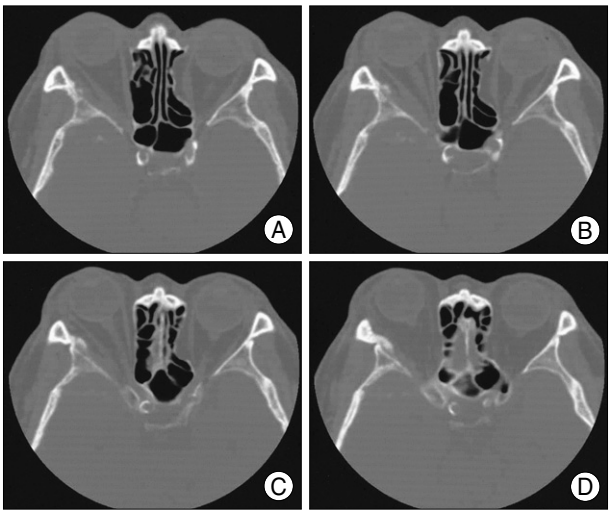

Fig. 2. Axial computed tomography scans show irregular bony destruction with sclerotic change of the right sphenoid wing.

was fragile because of tumor invasion. The dura was opened, and the involved portion was completely removed. The intradural tumor was partially adhered to the brain surface and it was totally removed. An artificial dura was used to close the dura, and the mesh and methylmethacylate were used to cover the bone defect. Histopathological examination revealed meningothelial meningioma without cellular atypia, mitosis, or anaplasia (Fig. 3).

Postoperative recovery was uneventful, and follow-up MRI 6 months after the surgery did not show evidence of recurrence.

DISCUSSION

MEP, a relatively uncommon tumor, is found in 2.5% of meningioma cases[3]. MEP is a tumor of limited thickness that grows along the planes of the meninges and, in some cases, occupies a considerable area[2,24]. It is found almost exclusively in middle-aged females whose symptoms are attributable to bony hyperostosis[8,14,26]. The spenoid ridge is the most common site of MEP[2,6,8,12,14,17,18], and insidious proptosis and associated ocular disturbances are the most common symptomatic presentations in patients with MEP of sphenoid ridge[3,8,14,26]. Proptosis is usually unilateral, non-pulsating, and irreducible, causing forward displacement of the eyeball[3].

Hyperostosis of the skull is one of the characteristic signs in MEP. It is observed frequently in MEP with an incidence of 13 to 49%, whereas it is seen in only 4.5% of all type of meningioma cases[12,17]. Kim et al.[18] classified 4 different patterns of hyperostosis in MEP according to CT features : 1) a homogenous pattern; 2) a periosteal pattern; 3) a three-layer pattern; and 4) a diploic pattern. The exact mechanism

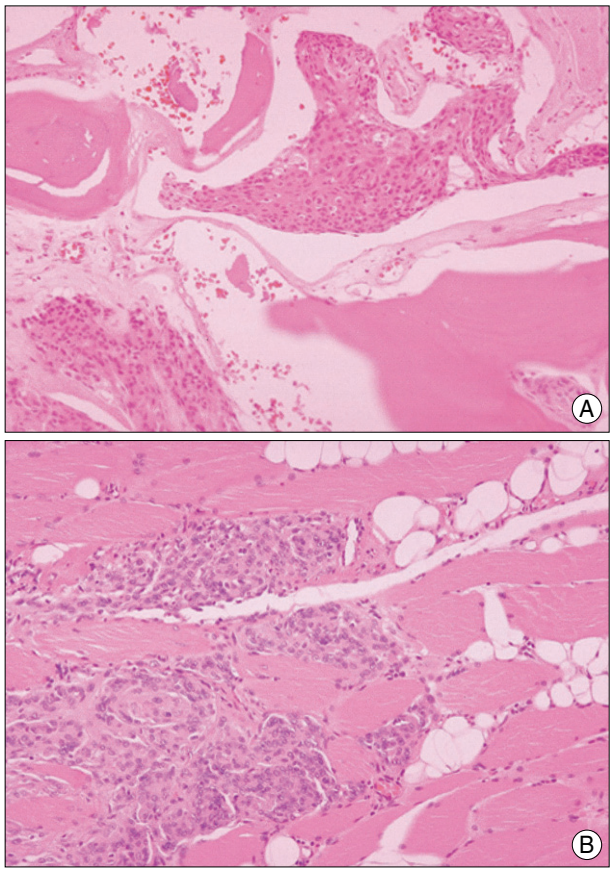

Fig. 3. A : Photomicrographs of a section of a biopsy specimen obtained from the right sphenoid wing. Tumor cells are arranged in a sheet or lobular configuration, and individual cells have round nuclei with inconspicuous nucleoli and indistinct cytoplasmic borders. They are infiltrating the surrounding bone in a sheet-like invasion pattern (H&E×100). B : Photomicrographs of a section of a biopsy specimen obtained from the right temporalis muscle. Tumor cells are dissecting to surrounding muscle bundles and adipose tissue. Tumor cells also show also ovoid nuclei and indistinct cytoplasmic borders, forming sheets or lobules (H&E×100).

of hyperostosis in MEP remains unclear. Several theories have been proposed, including vascular disturbance of the bone caused by a tumor, irritation of the bone caused by a tumor without invasion, stimulation of osteoblasts in normal bone caused by factors secreted by tumor cells, production of bone by the tumor itself, and tumor invasion of the bone[25]. Most authors agree with Cushing's[6] conclusion that the infiltration of the bone by meningioma cells stimulates osteoblastic activity, which result in hyperostosis[11,12,19,26,30].

Osteolysis in meningiomas is rare. Some authors believe that osteolytic meningiomas have malignant features and that osteolysis in meningiomas indicates a poor prognosis[4,15,23,31]. Russel and Rubinstein[28] have pointed out that meningiomas that present with osteolysis are more aggressive than others. Taveras and Wood[29] have proposed that purely osteolytic or destructive changes are more often associated with meningiomas that are primarily sarcomatous. Some authors

have reported osteolytic ectopic meningiomas[21,22,27] which are referred to as intraosseous meningioma[1,5,16], calvarial meningioma[20], or intradiploic meningioma[13]. They suggest that only a meningioma originating from the outer layer of the dura can evoke osteoclastic activity in the skull by causing mechanical pressure and erosion.

In our case, the tumor was an osteolytic MEP extending from the sphenoid wing into the middle fossa, the orbital wall and temporalis muscle. However, there was no evidence of malignancy according to histologic examination.

CONCLUSION

A case of osteolytic MEP of the sphenoid ridge was presented. This case merits attention due to its rarity, but more studies are necessary to determine the mechanism of osteolysis in MEP, which has no malignant features and is not an ectopic meningioma.

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