Hyperostotic Meningioma With Minimal Tumor Invasion Into the Skull
—Case Report—

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Abstract

A 56-year-old man presented with a meningioma associated with hyperostotic bone containing little tumor cell infiltration. The patient presented with a growing mass on his right forehead and exophthalmos. Computed tomography (CT) taken 4 years previously revealed only hyperostosis without intracranial lesion. Repeat CT revealed an enhanced intracranial mass with overlying diffuse hyperostosis extending extracranially. The tumor and affected bone were widely removed. Histological examination confirmed rhabdoid meningioma in the intracranial and extracranial lesion. However, most of the hyperostotic bone showed no tumor cell infiltration. The cause of hyperostosis associated with meningioma is unclear, but tumor invasion is the generally accepted cause. In this case, hyperostosis occurred without tumor cell infiltration so another mechanism was probably involved. The extracranial extension occurred despite the disproportionately small tumor without global tumor cell infiltration of the bone or bony erosion.

Key words: hyperostosis, tumor cell infiltration, extracranial extension

Introduction

Hyperostosis is a well-known sign of meningiomas, which is observed in 4.5% of all types, but is more frequently observed in meningioma en plaque with an incidence of 13% to 49%. Primary intraosseous meningioma induces hyperostosis in 60% of cases. The cause of hyperostosis is unclear, but tumor invasion appears to be generally accepted. We present a case of diffuse hyperostotic meningioma extending into the paranasal sinuses without evidence of tumor cell infiltration in most parts of the hyperostotic bone.

Case Report

A 56-year-old man presented with a growing mass on his right forehead and exophthalmos of the right eye persisting for 4 years in June 2004. He had attended another institution as a result of head trauma 4 years previously. Computed tomography (CT) demonstrated no specific findings. Since then, his symptoms gradually worsened but he was not concerned.

On admission, physical examination revealed an egg-sized lump on his right forehead above the eyebrow. The lump was firm with no tender points. In addition, the right eyeball protruded compared with the left. Neurological examination found no abnormalities. All laboratory results including the alkaline phosphatase level were all within the normal range.

CT taken in 2000 had revealed a sclerotic bony lesion slightly expanding the right frontal bone and the orbital roof, and a small soft tissue mass in the right frontal sinus and the ethmoid sinus, but no specific intradural or intracranial lesions (Fig. 1). After 4 years, CT in June 2004 revealed extensive progression of the lesion, and increased thickness and extent of the hyperostotic bony lesion (Fig. 2A). In addition, an enhanced intracranial lesion was observed, which had not been noted 4 years before. Preoperative magnetic resonance imaging revealed a more detailed appearance of the above findings (Fig. 2B, C).

A right frontal craniectomy was performed using a bicoronal skin flap. The thickened calvarium was...
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hard but slightly softer than the normal bone. The thickened frontal bone, including the frontal base with a normal bone margin, was widely removed. The underlying dura had adhered to the calvarium at focal areas and was thickened. Incision of the dura exposed a pinkish, friable, and soft mass, suggestive of meningioma, in the subdural space. The subdural mass including the thickened dura was removed. The frontal sinus was identified and the soft tissue mass within was removed. The slightly yellowish and soft mass in the ethmoid sinus was also removed through the frontal sinus. Macroscopically, there were no specific abnormal findings including erosive changes at the orbital roof and the frontal base. After the orbital roof was removed, the underlying periorbital yellowish mass was incised and removed. The orbital roof was reconstructed and the temporalis muscle flap was used to reinforce the anterior skull base. Cranioplasty was then performed. The postoperative course was favorable and he was discharged on postoperative Day 8.

Histological examination showed the tumor consisted of variable whorls and sheet-like growth of large cells with abundant eosinophilic cytoplasm, prominent eccentrically placed nuclei, and paranuclear inclusions (Fig. 3). Part of the tumor in the paranasal sinus also showed regions of fibroblastic meningioma. The pericranial soft tissue mass on the right forehead contained reactive and inflammatory tissue. Based on the above findings, the diagnosis was rhabdoid meningioma. However, most of the hyperostotic bone showed no tumor cell infiltration except for one tiny specimen (Fig. 4). Immunohistochemical examination showed the tumor expressed epithelial membrane antigen and myoglobin. The Ki-67 labeling index was <1%. Mitoses were scarce.

Discussion

The precise mechanism of hyperostosis associated with meningioma remains unclear. Many hypotheses regarding the mechanism of hyperostosis have been proposed, including prior trauma, vascular disturbance, irritation of the bone by the tumor without invasion, stimulation of osteoblasts...
tumor infiltration in our case. Two types of skull bone reactions to meningioma have been reported, osteoblastic and osteolytic changes.\textsuperscript{12} Osteoblastic changes are fairly common as a reaction to meningiomas, and there is usually little tumor cell invasion.\textsuperscript{4,12}

The histological type of meningioma appears to have no relationship with hyperostosis, but the various histological types are found as meningiomas en plaque, which are frequently associated with hyperostosis. In addition, there is no relationship between the hyperostotic pattern and the histological type of meningioma.\textsuperscript{7} Therefore, the rhabdoid type in our case probably did not influence the occurrence and pattern of hyperostosis. Rhabdoid meningiomas are quite rare and account for only 0.004\% of all meningiomas.\textsuperscript{2,9} The rhabdoid morphology is considered to indicate malignancy regardless of the mitotic index.\textsuperscript{2} The route of the extracranial extension in our case is unclear. The tumor could have extended into the paranasal sinuses without definite tumor cell infiltration and erosive changes in the skull base. The extension occurred from a relatively small intracranial tumor that was undetectable by CT 4 years previously. Intracranial meningiomas extend extracranially along well-recognized avenues: 1) by way of the neural foramina in the base of the skull, 2) along preformed pathways and spaces such as the sutures in the calvaria and the pneumatic cells in the temporal bone, and 3) through the skull by direct erosion.\textsuperscript{5,8} All three avenues were possible in our case, but the first avenue is the most likely because of the scant tumor cell infiltration of the bone and the absence of erosive changes in the skull base.

Hyperostosis associated with meningiomas usually originates from the infiltration of tumor cells. However, as our case shows, not all hyperostosis may originate from tumor cell infiltration, so other mechanisms may be involved. A small underlying intracranial tumor undetectable by CT can induce disproportionately great hyperostosis and extracranial extension. Extracranial extension may progress without definite tumor cell infiltration of the bone or bony erosion. However, tumor cell infiltration can only be excluded by extensive histological study. Wide surgical resection of the hyperostotic bone is the optimum treatment.

References


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