Anaplastic Meningioma With Extremely Rapid Recurrence
—Case Report—

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Abstract

A 62-year-old woman presented with an uncommon case of anaplastic meningioma manifesting as recent memory disturbance. Magnetic resonance imaging revealed a mass located in the right temporal lobe. She became unconscious because of uncal herniation and underwent urgent surgery. The tumor was completely resected, except for a lesion tightly attached to arteries. Histological examination indicated the presence of anaplastic meningioma with an extremely high MIB-1 labeling index (70%). After 43 days, the patient developed local recurrence and dissemination in the left temporal lobe. The exceptionally high MIB-1 labeling index corresponded with a short tumor doubling time (8.2 days). Whole-brain irradiation and linear accelerator surgery for disseminated lesions were performed, and the tumor growth halted. Although meningiomas rarely show malignant behavior, corresponding to World Health Organization grade III, it is necessary to consider malignant behavior when treating meningiomas.

Key words: anaplastic meningioma, recurrence, MIB-1, doubling time, radiation

Introduction

Meningiomas are common tumors arising from the meninges of the brain and spinal cord. Meningiomas constitute 30% of all primary brain tumors,9) and 90% are histologically benign and correspond to World Health Organization grade I.5) A few meningiomas, classified as atypical (grade II) and anaplastic (grade III), show aggressive clinical and histological features. We present a rare case of anaplastic meningioma that showed extremely rapid recurrence and intrathecal dissemination during the early clinical course.

Case Report

A 62-year-old woman with recent memory disturbance visited a local hospital. Magnetic resonance (MR) imaging revealed a mass located in the right temporal lobe, appearing as hypointense on T1-weighted imaging and isointense on T2-weighted imaging (Fig. 1A, B). T1-weighted MR imaging with gadolinium revealed a heterogeneously enhanced mass (diameter, 50 mm) with irregular margins (Fig. 1C, D). The tumor was associated with severe perifocal edema, causing midbrain compression. The patient was urgently admitted to our hospital because of left hemiparesis. She became unconscious with right mydriasis due to uncal herniation 2 days later. She underwent urgent surgery for tumor resection and external cranial decompression via a right frontotemporal approach. The tumor was completely resected, and the dura was coagulated at the sphenoid ridge, which was the tumor origin, except for the part surrounding the right middle cerebral artery, to which the tumor was tightly attached (Simpson grade III).

Histological examination revealed that the meningioma was malignant, with patternless growth, geographic necrosis (Fig. 2A), and no rhabdoid or papillary features. The meningioma fulfilled the criteria for anaplastic meningioma with \( \geq 20 \) mitoses per 10 high power fields (HPFs) (Fig. 2B). Immunohistochemical analysis showed partially positive staining for epithelial membrane antigen (EMA) and vimentin (Fig. 2C). The MIB-1 labeling index was 70%, determined as the percentage of positive nuclei from regions of maximal nuclear staining among 1000 tumor cells (Fig. 2D). Tumor cells were negative for desmin and silver impregnation, indicating absence of sarcoma (data not shown). The postoperative course was uneventful, and the mydriasis and gait disturbance immediately disappeared.

Postoperative MR imaging revealed subtotal removal of the tumor (Fig. 3A). However, MR imaging performed 43 days after the operation revealed tumor recurrence (size, \( 41 \times 39 \times 37 \) mm) and dissemination in the left temporal lobe (Fig. 3B). The tumor grew aggressively, and MR imag-
Fig. 1 A: T1-weighted magnetic resonance (MR) image showing a hypointense lesion in the right temporal region. B: T2-weighted MR image showing an isointense lesion. C, D: T1-weighted MR images with gadolinium enhancement showing a heterogeneously enhanced mass with irregular margins (diameter, 50 mm).

Fig. 2 A, B: Photomicrographs of the specimen obtained at the first operation showing atypical features, including patternless growth and geographical necrosis (A), and frequent mitoses (>20 mitoses per 10 high power fields; arrows in B). Hematoxylin and eosin stain, original magnification A: ×40, B: ×400. C: Immunohistochemical analyses showing partially positive staining for epithelial membrane antigen. Original magnification ×200. D: MIB-1 labeling index was 70%. Original magnification ×200.

Fig. 3 A: T1-weighted magnetic resonance (MR) image with gadolinium showing subtotal removal of tumor. B: T1-weighted MR image with gadolinium obtained 43 days after the operation showing dissemination in the left temporal lobe (arrow) and tumor recurrence (size, 41 × 39 × 37 mm). C: T1-weighted MR image with gadolinium obtained 55 days after the operation (tumor size, 54 × 53 × 57 mm) showing the diameter of the disseminated lesion is 7.0 mm (arrow). D: T1-weighted MR image with gadolinium obtained 64 days after the operation showing complete resection of the tumor and growth of the disseminated lesion (diameter, 9.7 mm; arrow).

Reoperation was performed, and the tumor was completely removed (Simpson grade II). Histological findings including immunohistochemical findings of EMA and MIB-1 were similar to those of the first operation. The disseminated lesion also showed extremely rapid growth, with the diameter increasing from 7.0 mm to 9.7 mm in 9 days (Fig. 3D). Whole-brain irradiation was performed (3 Gy × 12) immediately after the operation for the dissemination. Continuous linear accelerator surgery (18 Gy) was performed for the disseminated mass lesions. The tumor growth halted for 1 month.

Discussion

Anaplastic meningiomas constitute 1–3% of all meningiomas.9 The prognosis is poor despite surgical resection, as recurrence rates of anaplastic meningiomas are 50–80%, median survival is <2 years,9 and median time to recurrence is 9.6–42.1 months.1,5,10 In the present case, disturbance of unconsciousness rapidly progressed, and we had to operate urgently. The time to recurrence was 43 days, and doubling time was 8.2 days based on the MR imaging findings (Fig. 3B, C) performed using the formula described previously.11 Furthermore, the disseminated lesion in the left temporal lobe (Fig. 3C, D) showed a shorter
doubling time (6.4 days). This was an extremely aggressive anaplastic meningioma that showed very short doubling time.

Anaplastic meningiomas have histological features of malignancy with high mitotic index (>20 mitoses per 10 HPFs) in addition to the features of atypical meningiomas. Therefore, for example, Ki–67 proliferative index predicts clinical outcome in patients with atypical or anaplastic meningioma. In the present rare case of anaplastic meningioma showed extremely rapid growth, very short doubling time, and high MIB-1 labeling index, suggesting that malignant behavior must be considered when treating meningiomas.

Surgery is the primary treatment for all meningiomas, enables a definitive diagnosis, and extent of surgical resection is a significant factor for predicting outcomes in patients with meningiomas. Meningiomas should be totally removed including the site of dural origin (Simpson grade I). In this case, the first operation resulted in Simpson grade III due to the tight attachment of tumors to vessels, which might affect the rapid regrowth of the tumor. However, the role of surgery is less clear in anaplastic meningiomas than in benign and atypical meningiomas. Surgery is occasionally impracticable in anaplastic meningiomas because of multifocal recurrence and short recurrence interval. Further, surgery was not a significant prognostic factor based on univariate analysis of overall survival and progression-free survival. The extent of surgical resection was not correlated with the onset of recurrence. In our case, surgery was of limited use. We should have considered the next effective therapy immediately after the first operation.

Disseminated meningioma is mainly caused by malignant meningioma. Meningiomas arise from arachnoid cells and are exposed to the cerebrospinal fluid (CSF), but dissemination through the CSF is rare. Dissemination might be caused by surgery, but only surgery was not the cause of the dissemination because malignant meningiomas could spread without prior surgery. The cause of dissemination is still not clearly understood. In our case, the dissemination was presumably due to the operation because it appeared after the first operation in which the tumor was not removed entirely.

Generally, radiotherapy is performed for recurrent or residual meningiomas. Many studies have suggested radiotherapy for treating anaplastic meningioma, regardless of the extent of surgery. In our case, the tumor size remained stable for at least 1 month after radiotherapy. Considering the rapid tumor growth rate, radiotherapy was thought to be effective.

The present rare case of anaplastic meningioma showed extremely rapid growth, very short doubling time, and high MIB-1 labeling index, suggesting that malignant behavior must be considered when treating meningiomas.

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