Original article

Aggressive multiple lung metastases from intracranial atypical meningioma

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Abstract

Meningioma is usually benign, and extracranial metastasis from an intracranial meningioma is very rare. We discuss the clinical, radiological and histopathological presentation of an elderly man with pulmonary metastases from atypical meningioma (WHO grade II) . The patient was a 60-year-old male with aggressive pulmonary and intracranial metastases. However, there was no recurrence observed at the primary site treated by surgery and post-operative irradiation. The pulmonary metastases progressed rapidly, causing symptoms of respiratory failure, and the patient died 2 years after the initial treatment.

Key words: atypical meningioma, pulmonary metastasis, aggressive clinical course

Introduction

The prognosis of meningioma is generally favourable, being associated with the potential for cure with good quality of life. Although meningioma is usually benign, meningiomas are occasionally aggressive reducing the duration of survival. Such lesions include chordoid, clear cell, atypical, papillary, rhabdoid, and anaplastic meningiomas[1] . Despite complete resection, local recurrence has been noted in 9 to 32% of such cases[2] .

However, metastatic meningiomas are rare[3] , and have been estimated to occur at fewer than 0.1% of patients[2) 4)] . The mean interval from detection of the primary tumor to detection of the first metastasis is reported to be 6.4 years[3] .

Atypical meningioma is generally thought to be an intermediate grade between the benign and malignant forms[5] . We describe a very rare case of atypical meningioma which metastasized extensively to the lung, and followed an aggressive clinical course.

Case Report

A 60-year-old man noted the insidious onset of neuralgia of the extremities. Four months later, he was referred to Saitama Medical Center with left hemiparesis. MRI of the head demonstrated a tumor with peritumoral edema in the right parietal region and mild midline shift on T1- and T2- weighted images (**Fig.1**) . A Gd-enhanced coronal MR image of the head showed an enhancing homogeneous tumor invading the skull (**Fig.2**) . The findings on chest X-ray and bone scintigraphy were normal. He underwent a parietal craniectomy and excision of the tumor. At surgery, the dura was penetrated by the tumor adjacent to the periosteum. Histopathological examination demonstrated an atypical meningioma which was grade II in the WHO classification. There was some preservation of poorly formed whorls and many nuclei with prominent nucleoli, and high cellularity (**Fig.3**) . The surgical margin was microscopically positive. Postoperative irradiation was delivered with parallel opposed portals and the total dose was 50 Gy in 25 fractions.

One year and 4 months later, he developed intracranial central nervous system metastasis. A Gd-enhanced MRI of the brain demonstrated a

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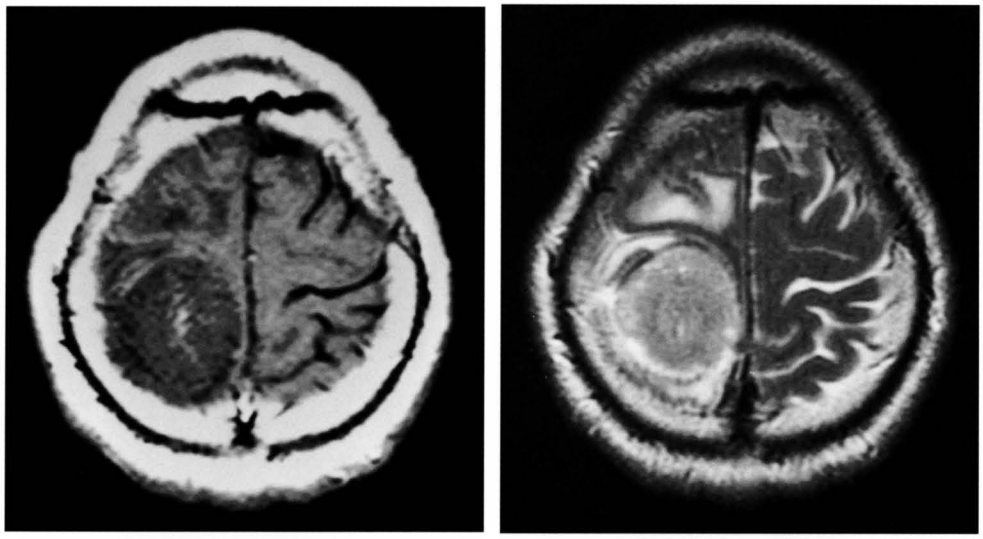

Figure 1. MRI of the brain demonstrated a slightly low intensity tumor in the right parieto-occipital region and mild midline shift on T1-weighted imege (Left) . T2-weighted image of the brain MRI showed a mild high intensity tumor (Right) .

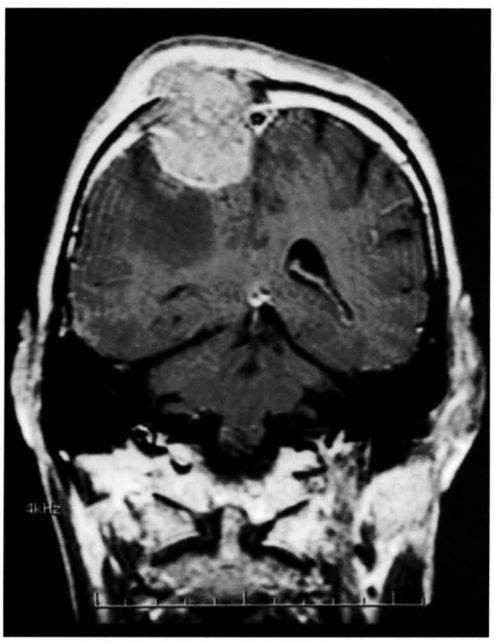

Figure 2. A coronal T1-weighted MR image obtained after Gd injection showed a dural-based enhanced tumor invading the skull.

nonhomogeneous enhancing metastatic tumor in the posterior fossa, although, there was no recurrence at the primary site. Radiation therapy was performed on the recurrent tumor at a dose of 50 Gy in 25 fractions with parallel opposed portals.

At that time, chest X-ray and CT scan showed

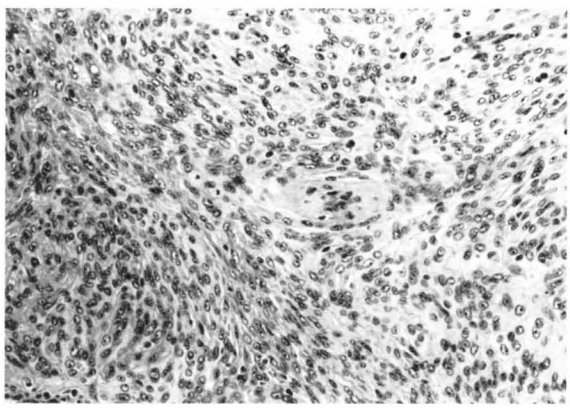

Figure 3. Photomicrograph of an atypical meningioma, showing poorly formed whorls, many nuclei with prominent nucleoli, and high cellularity.

multiple lung metastases. There was no mediastinal lymphadenopathy, pleural effusion, or other distant metastasis. Whole body ^{201}Tl image demonstrated abnormal accumulations in the intracranial metastasis and multiple lung metastases (**Fig.4**) . Over the subsequent 3 months, he developed dyspnea. Lesions of the lung metastases progressed rapidly, and pleural effusion appeared (**Fig.5**) . The patient died of lung metastases 2 years after the initial presentation. Although the histopathological diagnosis was atypical meningioma (WHO, grade II) , the patient had an aggressive clinical course.

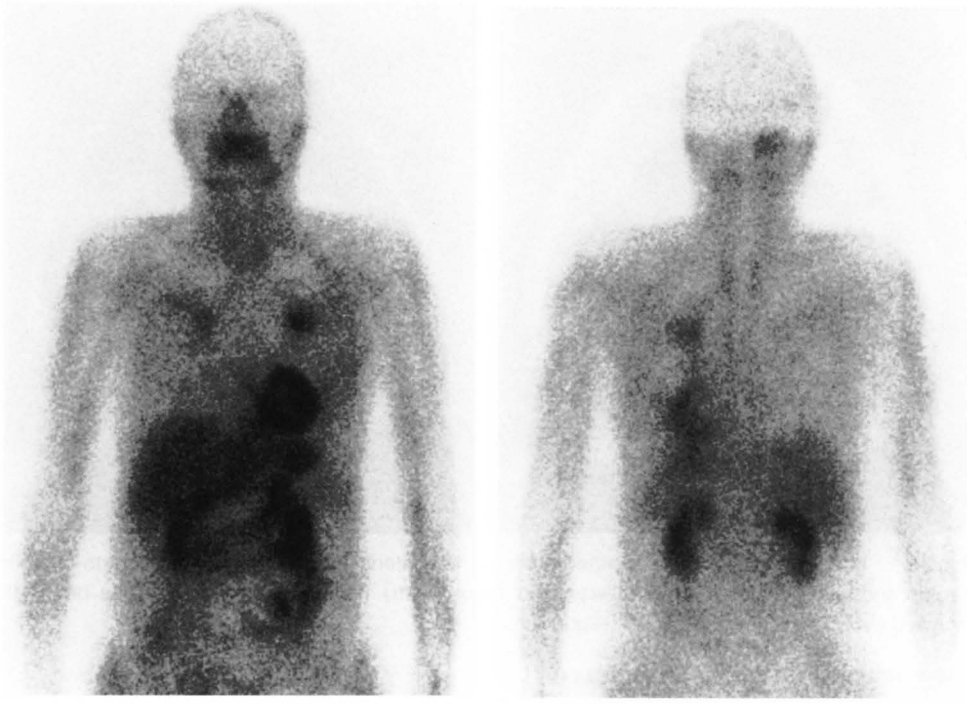

Figure 4. Whole body 201Tl scintigram showed abnormal accumulations in the intracranial metastasis and multiple lung metastases.

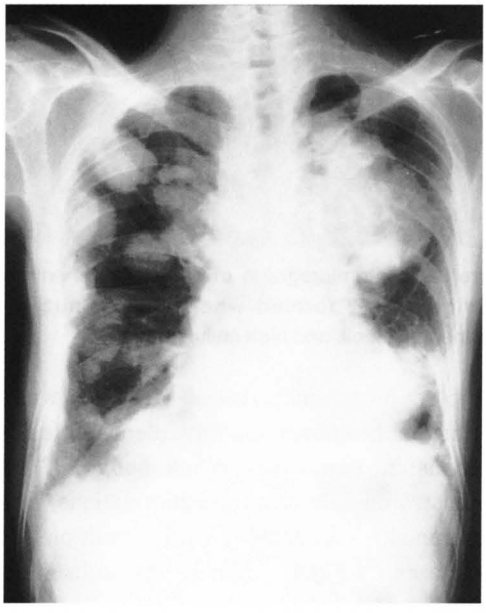

Figure 5. Chest X-ray showed multiple lung metastases that enlarged rapidly.

Discussion

Meningiomas are the most common non-glial intracranial tumors, representing 15 to 25% of all intracranial tumors.[1] However, metastasis of meningioma to distant extracranial sites is uncommon. Most patients with metastatic meningioma are adults between the ages of 40 and 60 years[2] . We presented the case of an elderly man with pulmonary metastases from an invasive and metastatic intracranial meningioma of atypical histology.

The histologic malignancy index is associated with the locally aggressive character and metastasis[6] . However, even when the histopathology shows malignant features, metastases are uncommon. The incidence of metastasis from this tumor is as low as 0.1%. Metastatic meningioma is not usually benign, however, a review by Tominaga et al.[4] found that more than 60% of reported extracranial metastases from meningioma were from benign meningiomas. Benign meningioma retains meningothelial whorls, does not usually invade the brain, and has only a small area of necrosis. However, aggressive meningiomas show the

areas of necrosis, increased cellularity, high nuclear/ cytoplasmic ratio, prominent nucleoli, and sheetlike growth[1] . Perry[7] reported that the histologic variables of the greatest prognostic significance were frank anaplasia, excessive mitotic index, and nuclear atypia. The histopathology of this patient showed high cellularity, and many nuclei with prominent nucleoli, mitosis, and poorly formed whorls.

Although extracranial metastases are rare, the lung, the abdomen, cervical lymph nodes and bones have been reported as the most common sites of metastasis from meningiomas[4] [8] . Hematogeneous metastasis of meningioma is probably most frequently the result of the occasional invasion of the venous sinuses and large vessels[4] . Our case had both intracranial metastasis and systemic metastases to the lung.

Complete surgical resection is the treatment of choice for accessible intracranial or intraspinal meningiomas. Postoperative radiation therapy is controversial, but it has been recommended for the prevention of local recurrence, especially when resection is subtotal or when the histology suggests malignancy[9] . Younis reported that despite treatment with either chemotherapy or radiotherapy, the prognoses of these patients do not improve[10] . Local recurrence of meningioma is usually difficult to control and increases the morbidity of the patient[11] . In this case, there was no recurrence at the primary lesion site treated with postoperative radiotherapy.

Stoller reported that pulmonary metastases only rarely become symptomatic and were sometimes detected only at necropsy[3] . Lung metastases present as single or multiple round non-calcified parenchymal nodules of varying sizes, and multiple deposits are noted in half of the cases[12] . LeMay reported a slow rate of growth for the lung nodule[11] . Drummond described that metastases of meningiomas were often asymptomatic and rarely caused death[13] . However, our patient developed dyspnea because the metastatic tumors of the lung grew rapidly. Although the histopathology showed an intermediate grade, the patient followed an aggressive clinical course. We reported a case of aggressive metastasis from atypical meningioma.

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