

A Rare Case of Femur Metastasis from Brain Meningioma

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Abstract

Background/Aim: Metastases of meningiomas are infrequent, and extracranial bone involvement is extremely rare. We describe a patient with femoral metastasis originating from an atypical brain meningioma.

Case Report: A 55-year-old male had undergone five surgical procedures and Gamma Knife® radiosurgery for brain meningioma over five years. Four months after the final treatment he presented with spontaneous right-hip pain. Radiography showed a radiolucent lesion at the lesser trochanter; computed tomography confirmed cortical bone destruction. Positron emission tomography–computed tomography demonstrated abnormal uptake in the femur [Standardized Uptake Value (SUV) 3.3] and in residual intracranial tumor (SUV 9.9). Magnetic resonance imaging revealed low T1- and high T2-weighted signal intensities at the lesion. Open biopsy identified metastatic WHO grade II meningioma positive for epithelial membrane antigen and somatostatin receptor 2A, with a Ki-67 labelling index of approximately 10%. Wide resection of the proximal femur with endoprosthetic reconstruction was performed. Postoperatively, the patient experienced intracranial tumor recurrence and received radiation therapy. No local recurrence or additional metastases were observed during the three-year follow-up; the Enneking functional score was 60%.

Conclusion: We report on an extremely rare instance of metastasis to the femur in a case of atypical brain meningioma.

Keywords: Atypical meningioma, femur, bone metastasis, femoral lesion, orthopedic oncology, case report.

Introduction

Meningiomas are the most common primary central nervous system neoplasms and constitute more than 30% of all intracranial tumors (1, 2). Most are benign; therefore,

extracranial metastases are infrequent, occurring in only 0.1% of cases (1-3). Approximately 6% of meningiomas are classified as World Health Organization (WHO) grade II (atypical), and their five-year survival rate is 68.8% (4, 5). The most frequent site of extracranial metastasis is the



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lung, whereas bone involvement is rare (6-8). We report an extremely rare instance of metastasis to the femur in a patient with atypical (grade II) meningioma.

Case Report

A 55-year-old male with a history of multiple surgeries and Gamma Knife radiosurgery for brain meningioma over five years presented to our hospital with spontaneous right-hip pain, which began four months after his final treatment. On physical examination, there was no redness or local heat, but tenderness and hip motion pain were present. Radiographic examination (X-ray) revealed a radiolucent area at the lesser trochanter (Figure 1). Computed tomography (CT) showed cortical bone thinning and destruction at the lesser trochanter (Figure 2). Positron emission tomography-computed tomography (PET-CT) demonstrated abnormal uptake in the brain meningioma and right proximal femur with a Standardized Uptake Value (SUV) of 3.3 at the lesser trochanter and 9.9 at the meningioma (Figure 3). The Mirels score (9), used to assess the risk of pathological fracture, was 9 (Site: 3, Nature: 3, Size: 1, Pain: 2). Magnetic resonance imaging (MRI) showed low signal intensity on T1-weighted images and high signal intensity on T2-weighted images at the lesser trochanter (Figure 4, Figure 5). An open biopsy confirmed the diagnosis of femur metastasis originating from a grade II brain meningioma. Histopathological analysis revealed the proliferation of tumor cells resembling meningocytes in a sheet-like or partially whorled pattern (Figure 6). The nuclei of the tumor cells were oval to round, with nuclear enlargement and pleomorphism. Mitotic figures were observed at approximately 1 per high-power field (HPF, $\times 400$) (Figure 7). Immunohistochemistry showed that the neoplastic cells were positive for epithelial membrane antigen (EMA) and somatostatin receptor 2A (SSTR2A), and the Ki-67 labeling index (L.I.) was approximately 10%. We performed wide resection of the tumor and prosthetic replacement of the femur. Postoperatively, the patient experienced recurrence in the brain meningioma and

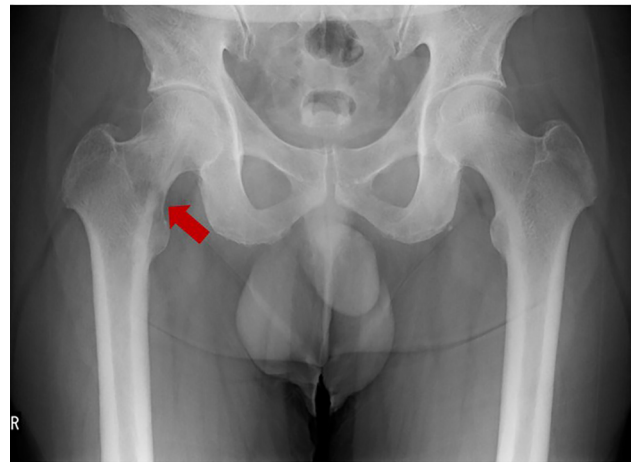


Figure 1. Plain X-ray showing a radiolucent area at the lesser trochanter.

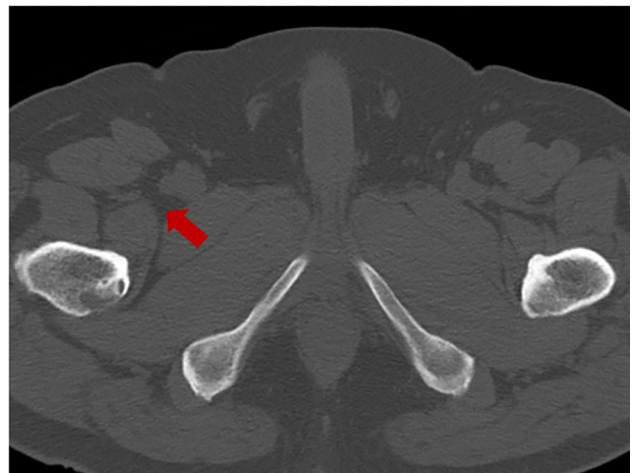


Figure 2. Computed tomography image showing cortical bone thinning and destruction at the lesser trochanter.

received radiation therapy. However, there was no local recurrence or metastasis observed in other sites except brain 3 years postoperatively, and the Enneking functional evaluation score (10) was 60%.

Discussion

Meningioma is one of the most common benign tumors of the central nervous system. Extracranial metastases from

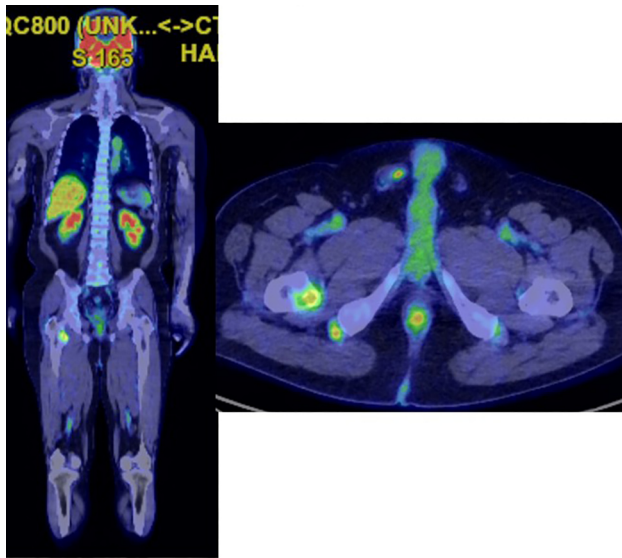


Figure 3. Positron emission tomography-computed tomography demonstrating abnormal uptake with a Standardized Uptake Value (SUV) of 3.3 at the lesser trochanter and 9.9 at the brain meningioma.

meningiomas are very rare (1-3). The most common metastatic site is the lung, followed by the liver and regional lymph nodes, while bone is considered a rare site of metastasis (6-8). Spinal and sacrum metastases are the most frequently reported bony metastases (6-8, 11-15). To the best of our knowledge, there have been no report of metastasis to the femur without other metastasis, as seen in this patient.

Multiple treatments for local recurrence may cause metastasis of atypical meningioma (16). The interval between the first treatment and metastasis to the bone can vary from three to 168 months, indicating a wide range in the time course of metastatic progression in patients with meningioma (17, 18). Previous reports indicate that the interval is shorter in patients with high-grade meningioma (grade II, III) (mean 24.5 ± 16.3 months) compared to those with low-grade meningioma (grade I) (mean 98.3 ± 12.7 months) (16, 19). Recently, we experienced metastasis to the sternum from the atypical meningioma (grade II) in a patient who underwent surgery and gamma-knife treatment for grade II brain meningioma over 31 years (20). Although the metastatic progression of high-grade

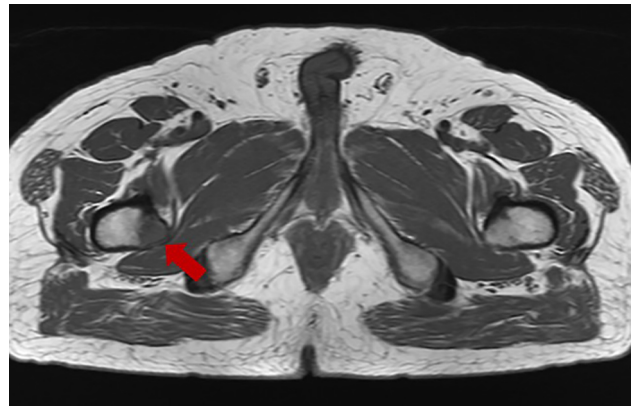


Figure 4. T1-weighted magnetic resonance imaging showing low signal intensity at the lesser trochanter.

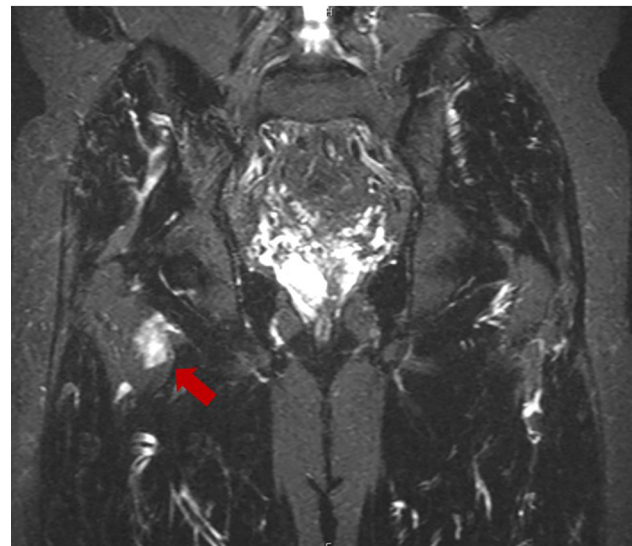


Figure 5. T2-weighted magnetic resonance imaging showing high signal intensity at the lesser trochanter.

meningioma varies significantly across reports, the duration of this case was extremely long compared to the typical time course. Therefore, long-term follow-up may be necessary to identify metastases even in patients with high-grade meningioma. In this case, the patient underwent surgery and gamma-knife treatment for grade II brain meningioma over 5 years and suffered from spontaneous right hip pain four months after the final

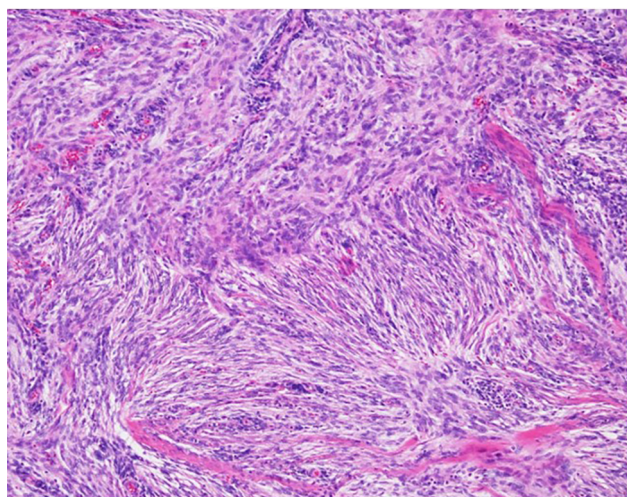


Figure 6. Histopathological specimen showing tumor cells resembling meningiocytes in a sheet-like or partially whorled pattern (hematoxylin-eosin, $\times 100$).

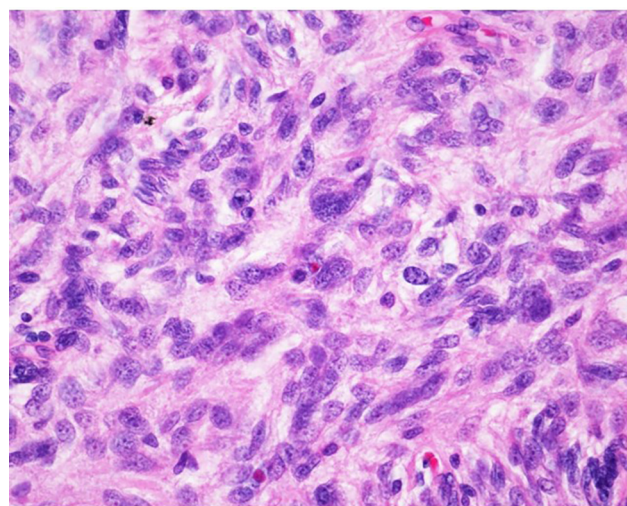


Figure 7. High-power view of the histopathological specimen showing oval to round nuclei with nuclear enlargement and pleomorphism; mitotic figure visible (hematoxylin-eosin, $\times 400$).

treatment. It took approximately five years from the start of treatment to the onset of metastasis, which is considered relatively long for a grade II meningioma.

The treatment for high-grade meningioma involves surgical resection and focal radiation, depending on the complexity of the individual case. There is no established

therapy for metastatic meningiomas (21, 22). The role of chemotherapy for recurrence remains inconclusive in the literature (15, 23). Although radiotherapy and chemotherapy are sometimes used for treating metastases of brain meningiomas tumor resection was chosen in this case due to the risk of pathological fracture, according to Mirels score. In this case, bone metastasis occurred during long-term treatment for primary meningioma over five years. This case suggests that long-term follow-up of meningioma treatment may reveal potential for bone metastasis.

Conflicts of Interest

The Authors declare no conflicts of interest in relation to this study.

Authors' Contributions

YM, TM, OS, KN, MN, RM, KG and HT performed the surgeries and clinical treatments. TN, YM, TM, OS, KN, MN, RM, KG and HT analyzed the pathology and clinical course. YM, TM and TN took the lead in drafting the manuscript, with support from SO, KN, MN, RM, KG and HT. All authors discussed the clinical results, critically reviewed the report, commented on successive drafts and approved the final manuscript. NA supervised the findings of this case and helped to draft the manuscript.

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Artificial Intelligence (AI) Disclosure

No generative AI tools were used in the preparation of this manuscript.

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