The Ochsner Journal 15:200–202, 2015
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Benign Sacral Metastatic Meningioma: A Rare Entity

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Background: Meningiomas are common intracranial tumors with a low metastatic rate. Those that do metastasize often show histopathologic signs of malignancy. In rare cases, the primary and secondary tumors are histologically benign.

Case Report: We report the case of a 57-year-old female with a histologically benign intracranial meningioma that metastasized to the sacrum. The patient had a long history of intracranial meningioma with multiple recurrences. At each recurrence, histopathologic examination of the resected tumor showed no signs of malignancy. The sacral meningioma was biopsied and found to be histologically benign. The patient was treated with radiotherapy (54 Gy in 30 fractions), and her symptoms resolved. Six months later, the patient developed left leg weakness. Magnetic resonance imaging showed growth of her intracranial mass for which she underwent a craniotomy for tumor resection. Pathologic evaluation showed evidence of benign meningioma without atypical features. She recovered well from this procedure and returned to her baseline in several weeks.

Conclusion: After treatment, the patient had no signs of radiographic progression in either location.

Keywords: *Meningioma, neoplasm metastasis, radiosurgery, sacrum, spine*

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INTRODUCTION

Meningiomas are the most common intracranial tumor, comprising 13%-26% of all primary brain tumors.[1] Although very common, they rarely metastasize with a rate of <1 in every 1,000 meningiomas.[2] The most frequent locations for metastasis are lung and pleura, liver, lymph nodes, and bone.[3] Almost all cases that metastasize show histopathologic and/or biological signs of malignancy.

In rare cases, both the primary and secondary tumors are histologically benign. We report a case of benign intracranial meningioma metastasizing to the sacrum. A review of the literature resulted in only one previous report of a benign metastatic meningioma to the sacrum.[4] In that case, the patient had no histopathologic features of malignancy in either the primary or the metastasizing tumors.

CASE REPORT

A 57-year-old female with a 30-year history of a parietal convexity meningioma first underwent surgery in 1985 and had 7 subsequent surgeries including gamma knife radiosurgery to the intracranial tumor bed in May 2008. The initial operation achieved a macroscopically complete resection with a histologic classification of meningothelial meningioma. Pathologic analysis of the tumor from subsequent surgeries consistently demonstrated benign meningioma. The patient had a residual tumor ($1.5 \times 1.5 \times 1.8$ cm) that we followed with serial magnetic resonance imaging (MRI). She recently presented with lower back pain. MRI revealed a fracture of her sacrum with evidence of bone marrow replacement within an area of gadolinium enhancement in the left sacral area measuring 6.4×3.4 cm (Figure 1, A and B).

The patient underwent kyphoplasty to alleviate her back pain with concurrent biopsy of the sacral mass. Histopathologic analysis of the core biopsy of the sacral mass (Figure 2A) revealed whorl-shaped spindle cells typical of meningothelial meningioma. Atypical features, such as nuclear pleomorphism, necrosis, and mitosis, were not identified. Tumor cells from the sacral mass were positive for epithelial membrane antigen (Figure 2B) and progesterone receptor (Figure 2C) and negative for S-100 protein (Figure 2D). Based on the histopathologic findings, a radiosurgery treatment plan was selected. The patient underwent a course of radiotherapy at 1.8 Gy per fraction for a total of 45 Gy. Following the initial 45 Gy treatment, a boost course using intensity-modulated radiation therapy (IMRT) was delivered at 1.8 Gy per fraction for a total of 9 Gy. The purpose of treating the patient with a boost course using IMRT was to spare the small bowel from excessive amounts of radiation. No signs of radiographic progression were seen in the sacral area following radiotherapy (Figure 1, C and D).

Six months following completion of radiotherapy to the sacral area, the patient developed left leg weakness. MRI showed growth of her intracranial mass ($2.6 \times 2.8 \times 2.4$ cm) (Figure 3, A and B). She underwent a craniotomy for tumor

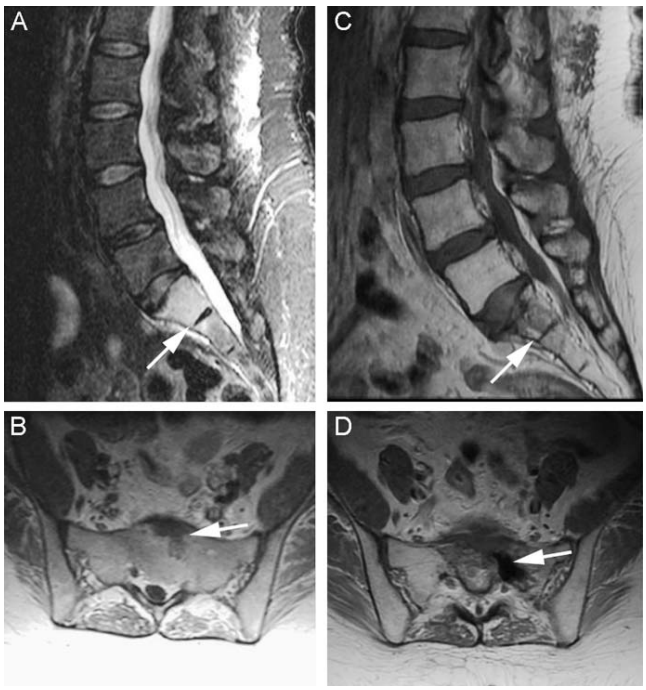

Figure 1. T1-weighted magnetic resonance images (MRIs) of the sacral lesion showing enhancement (arrow) in sagittal (A) and axial (B) views. Sagittal (C) and axial (D) T1-weighted MRIs of the sacral lesion after radiotherapy demonstrate no enhancement (arrow).

resection. Pathologic evaluation showed evidence of a benign meningioma without atypical features (Figure 4, A and B). She recovered well and returned to baseline in several weeks. No radiographic evidence of recurrence has been seen (Figure 3, C and D).

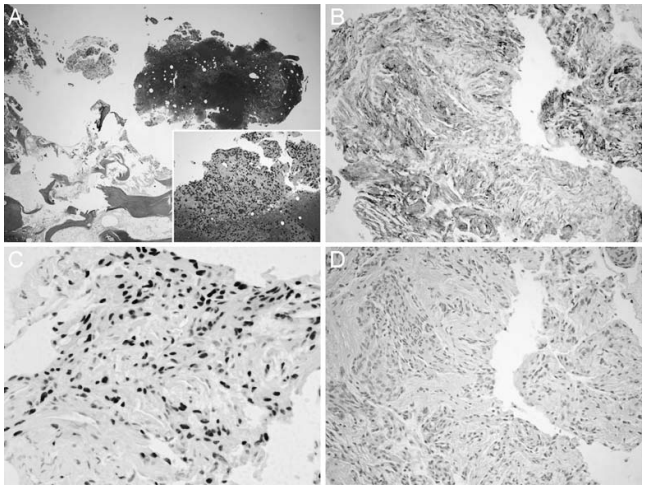

Figure 2. Photomicrographs of the sacral mass showing meningothelial meningioma (A), hematoxylin and eosin (H&E) stain, original magnification ×25 (H&E stain inset, original magnification ×200). No atypical features were observed. The tumor was positive for (B) epithelial membrane antigen (EMA) and (C) progesterone receptor and negative for (D) S-100 protein (EMA and S-100 protein original magnification ×200, progesterone receptor original magnification ×400).

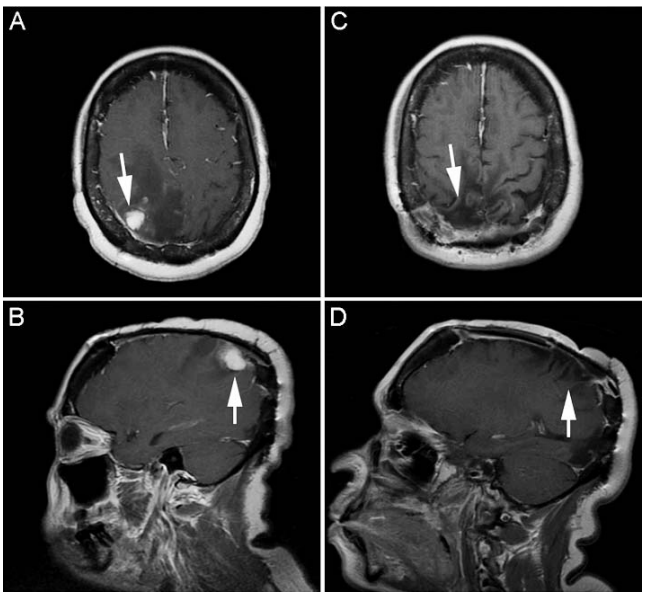

Figure 3. T1-weighted magnetic resonance images (MRIs) of the intracranial lesion showing enhancement (arrow) in axial (A) and sagittal (B) views. Axial (C) and sagittal (D) T1-weighted MRIs of the intracranial lesion after tumor resection demonstrate no enhancement (arrow).

DISCUSSION

Meningiomas are generally considered benign tumors. Given the frequency of these tumors, metastasizing meningiomas are a rare event. Surov et al reviewed the literature from 1990 to 2012 and reported only 115 cases with metastasizing meningiomas.[5] Interestingly, several authors have reported that roughly 60% of metastatic meningiomas arise from morphologically benign primaries.[2,5,6] Despite the high percentage of metastatic meningiomas arising from benign parent tumors, the majority of them show signs of malignancy. Only 25 cases have been reported in which both the primary and extracranial metastatic meningiomas were histologically benign.[4,7-10]

Spinal metastasizing meningiomas have been rarely reported in the literature, with only 19 cases reported to date[11] and only one previously reported case of sacral metastatic meningioma.[4] Similar to our patient, the histologic appearance of the primary and secondary tumors was benign. For our patient, the sacral mass was treated with radiosurgery, while in the previous case, the patient was treated with an anterior-posterior-anterior approach with

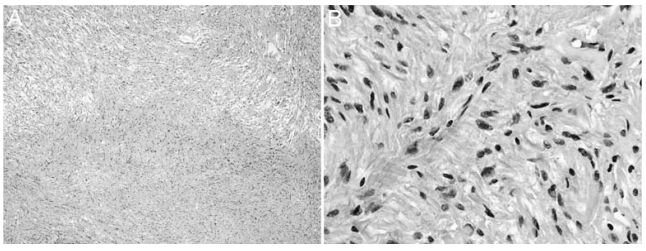

Figure 4. Photomicrographs of intracranial tumor showing meningothelial meningioma (A) hematoxylin and eosin (H&E) stain, original magnification ×50 and (B) H&E stain, original magnification ×400.

total excision of the tumor. Both patients appear to have tolerated treatment and were able to ambulate freely with improved clinical symptoms.

Several routes for the spread of meningiomas have been identified in the literature, including hematogenous, lymphatic, and cerebral spinal fluid routes.[2] Based on the previously observed localization of metastasis, the hematogenous route via the jugular vein may be the primary method of dissemination in meningiomas.[5] The paravertebral venous plexus has also been suggested to play a role in metastatic spread with involvement of the vertebrae, kidney and perirenal tissue, and adrenal glands.[5] Lymphatic and cerebrospinal fluid pathways may also provide a route for metastatic invasion.[12] Surov et al identified meningioma spreading via the cerebrospinal fluid pathway in 15.5% of all analyzed metastatic meningiomas.[5] The route of metastasis in our patient was likely through the cerebrospinal fluid pathway because the sacral region is the only known site of metastasis.

CONCLUSION

Our case is the second reported case of a benign sacral metastasizing meningioma. The sacral meningioma was treated with radiotherapy without any complications, and after treatment the patient had improved clinical symptoms with no signs of radiographic recurrence.

ACKNOWLEDGMENTS

This work was supported by a Ruth L. Kirschstein National Research Service Award F30CA177267 from the National Cancer Institute to Michael J. Strong. The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

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