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Case Report & Case Series

Benign metastasizing leiomyoma mimicking dumbbell tumor of the spine: A report of two cases

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ABSTRACT

Benign metastasizing leiomyoma (BML) is an extrauterine smooth muscle tumor that occurs in patients with a current or prior history of uterine leiomyoma. BML in the spine is extremely rare. We report 2 cases of spinal BML mimicking dumbbell tumors in the cervical or lumbar spine. Tumors in both cases relapsed after tumor resection; however, the tumor did not progress while on hormone therapy.

1. Introduction

Leiomyomas are benign tumors mainly occurring in the uterus. Benign metastasizing leiomyoma (BML) is an extrauterine smooth muscle tumor that occurs in patients with a current or prior history of uterine leiomyoma [1,2]. BML of the spine is extremely rare with only six reported patients to date [3–8].

2. Case report

2.1. Case 1

A 44-year-old woman developed numbness and radicular pain on the right arm 7 years after uterine resection for leiomyoma. Neurological examination revealed motor weakness and sensory disturbance on the right arm. Magnetic resonance (MR) examinations revealed that a dumbbell tumor was present along the spinal canal, right intervertebral foramen, C6 vertebral body, and paravertebral area at the C5–6 level (Fig. 1A). An anterior and posterior approach was performed for laminectomy, resection of tumor, and C6 corpectomy following instrumented spinal fusion. Histologic examination of the surgical specimen revealed a benign leiomyoma with spindle cell proliferation (Fig. 1B).

The recurrence of tumor and deterioration of right arm pain occurred 2 years after the operation. A lateral approach was performed for the resection of recurrence tumors. A second recurrence of tumor was revealed by MR examination 2 years after revision surgery.

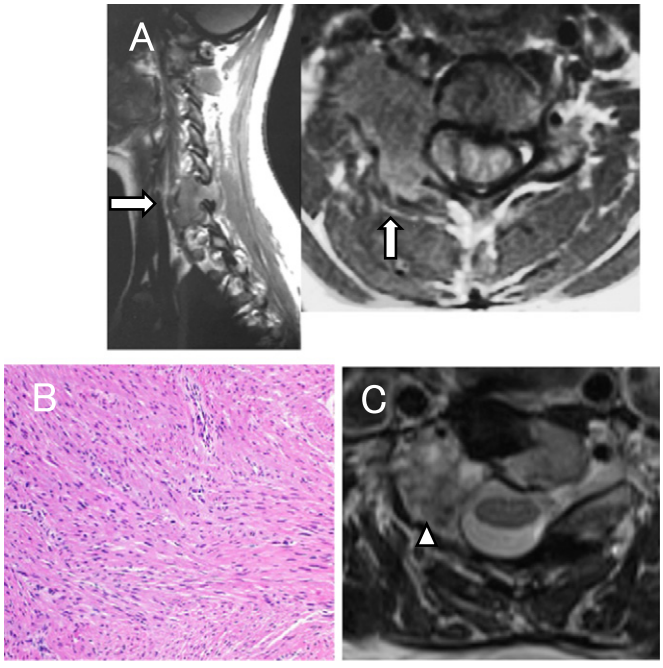

Fig. 1. Case 1: 44-year-old woman. A: Magnetic resonance (MR) examinations revealed that a dumbbell tumor was present along the spinal canal, right intervertebral foramen, C6 vertebral body, and paravertebral area at the C5–6 level (arrow). B: Histologic examination of the surgical specimen revealed a benign leiomyoma. C: The recurrent tumor was shrinking after hormone therapy (arrow head).

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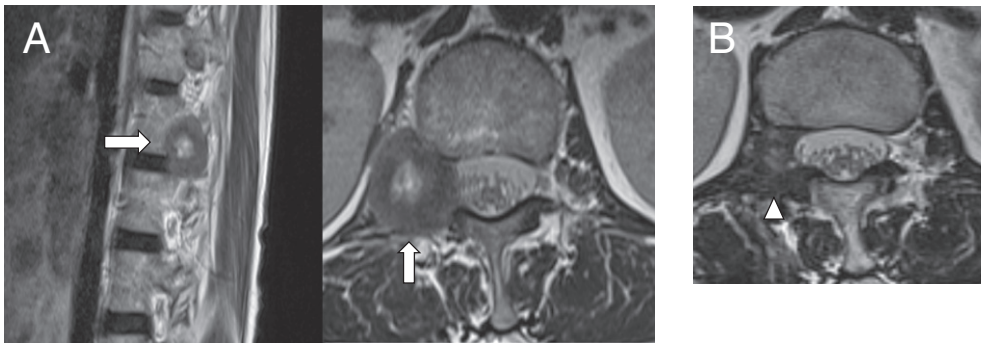

Fig. 2. Case 2: 59-year-old woman. A: MR examinations revealed that a dumbbell tumor was present in the right intervertebral foramen at the L1–2 level (arrow). B: The size of the recurrent tumor was maintained after hormone therapy (arrow head).

Recurrent tumors shrank and arm pain was resolved after hormone therapy using danazol (Fig. 1C). The hormone therapy has been continuing, and both tumor shrinkage and resolving of arm pain have been maintained for 15 years.

2.2. Case 2

A 59-year-old woman developed low back pain and received uterine resection for leiomyoma 1 year 3 months and, 1 year 2 months before admission, respectively. Neurological examination did not reveal any motor weakness or sensory disturbance. MRI revealed that a dumbbell tumor was present in the right intervertebral foramen at the L1–2 level (Fig. 2A). A posterolateral approach was performed for the resection of the tumor. Histologic examination of the surgical specimen revealed a benign leiomyoma (Fig. 3). The neoplastic cells immunoexpressed smooth muscle actin (Fig. 3B) and were negative to S100 protein, CD34. Recurrence of tumor was revealed by MR examination 8 months after surgery. The size of the recurrent tumor was maintained for 2 years with hormone therapy using letrozole (Fig. 2B).

3. Discussion

We present 2 rare cases of spinal BML mimicking dumbbell tumors in the cervical or lumbar spine. BML in the spine is very rare, and to our knowledge only 6 cases of BML in the spine have been reported in literatures [3–8]. Five of eight cases, including the present case, were in the cervical spine, and 2 of 8 were in the sacrum. One of the present cases (case 2) may be the first instance of BML reported in the lumbar spine. BML should be considered in the differential diagnosis of spinal tumors even if they are mimicking dumbbell tumors in patients who have a history of uterine leiomyoma.

Since uterine leiomyoma is a hormone-dependent tumor, hormone therapies have been proposed [2,5,7]. The tumors in both cases were relapsed after tumor resection; however, the tumor did not progress while on hormone therapy.

Conflicts of interest/Disclosures

The authors declare that they have no financial or other conflicts of interest in relation to this research and its publication.

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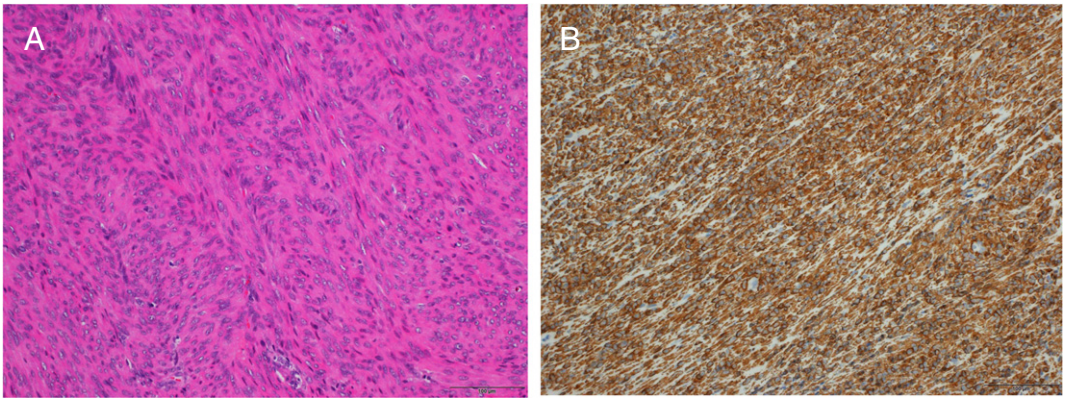

Fig. 3. A. Histologic examination of the surgical specimen revealed a benign leiomyoma with spindle cell proliferation. B: The neoplastic cells immunoexpressed smooth muscle actin.