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# Metastatic Spinal Epidural Leiomyoma: A Case Report[1]

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We report here on a case of a spinal extradural leiomyoma in a 67-year-old woman, and this tumor was in a very unusual location for a leiomyoma. Because the patient underwent hysterectomy for a uterine leiomyoma 20 years ago, we can speculate that the spinal lesion was a metastatic leiomyoma.

**Index words :** Spinal cord
Neoplasms
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Leiomyomas are benign tumors and they mainly occur in the uterus. Primary leiomyomas in the central nervous system are extremely rare (1). Primary spinal leiomyomas have only rarely been reported in patients with acquired immune deficiency syndrome (AIDS), and this is irrespective of the concomitant presence of uterine leiomyoma (2, 3).

Although this histologically benign tumor is a rare finding, it can metastasize to many locations, which is a well known phenomenon that's termed benign metastasizing leiomyomas (BMLs). The pathogenesis of metastasis has not been fully elucidated. However, women who have undergone hysterectomy for leiomyomas are most commonly affected (4 - 8), and the lung is the most common site of involvement (4).

We report here on a case of a lumbosacral, extradural leiomyoma in a woman who had a previous history of hysterectomy for treating her leiomyoma 20 years previously.

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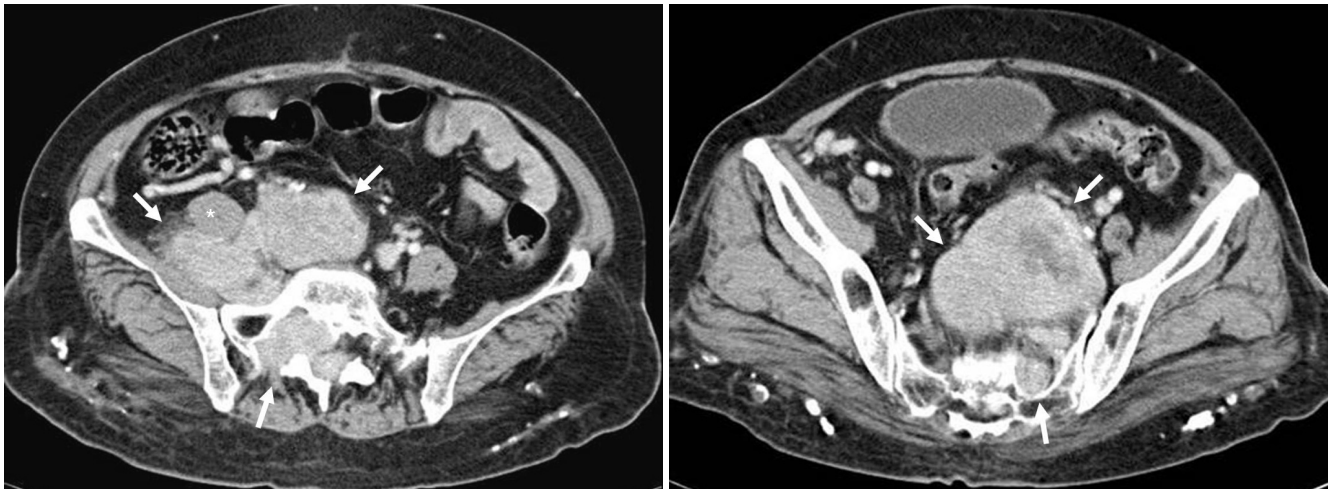
## Case Report

A 67-year-old woman presented in April 2004 to the neurosurgery department with a history of monoparesis of the left leg. Upon examination she exhibited a mild left lower extremity motor deficit (grade 4/5) without any sensory deficit.

She had undergone hysterectomy for leiomyoma 20 years prior to presentation. A contrast-enhanced computed tomography (CT) scan showed a lobulated, homogeneously enhancing soft tissue mass in the right S1 paravertebral region, the pelvic cavity and the spinal canal, and this was seen together with vertebral body erosion (Fig. 1A, B). The sagittal T1-weighted MR imaging demonstrated a soft tissue mass in the spinal canal from the L5 to S2 level and also in the prevertebral space (Fig. 2A). On the sagittal T2-weighted MR images, the lesions demonstrated nearly isosignal intensity to the adjacent muscles (Fig. 2B). The mass showed strong, homogeneous contrast enhancement on a gadolinium-enhanced T1-weighted MR image. A contrast-enhanced axial T1 image showed the intraspinal mass exiting the spinal

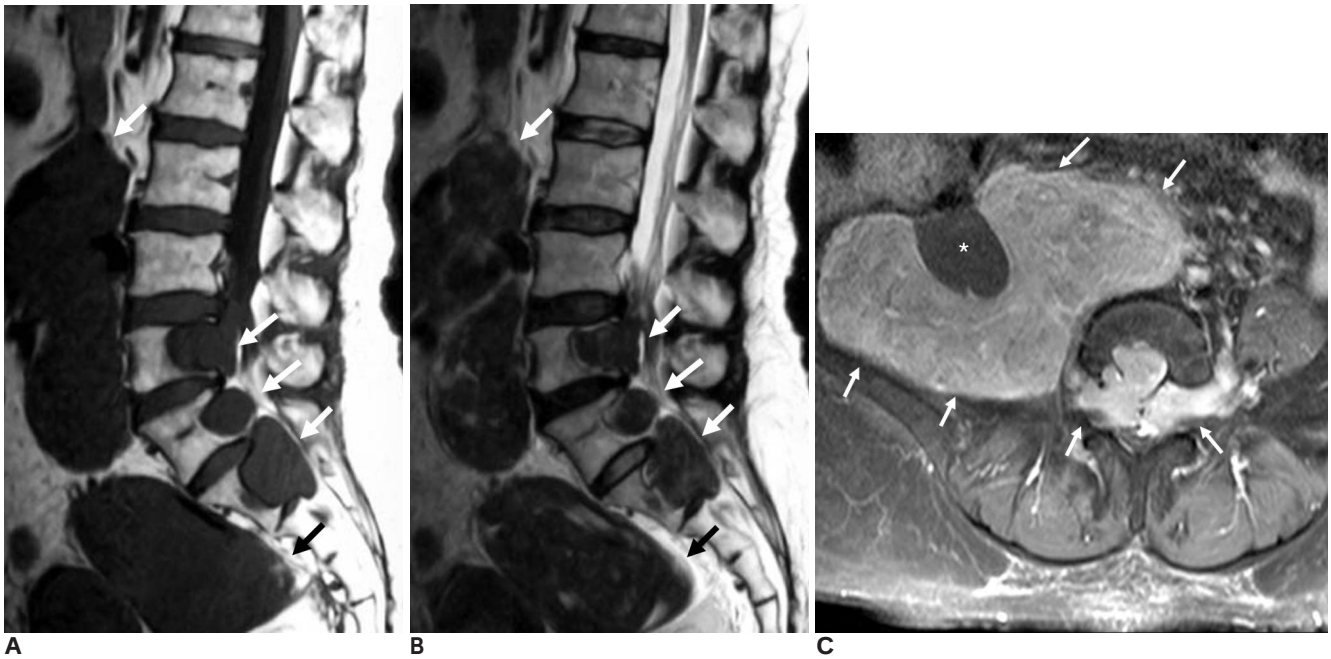
canal through the bilateral neural foramina at the L5 level. It formed a large bilobulated paraspinal component along the medial and lateral sides of the right psoas muscle, displacing it anterolaterally (Fig. 2C). Another large mass in the pelvic cavity (Fig. 1B) was connected with the intraspinal lesion through the left S2 neural foramen.

Although the T2 signal intensity of the tumor was not as bright as a typical neurofibroma, this was our initial radiologic diagnosis due to the lesion's characteristic imaging findings as described above. Total laminectomy at L5 and S1 with total excision of the tumor was performed. The lesion was intraspinal and extradural in location.



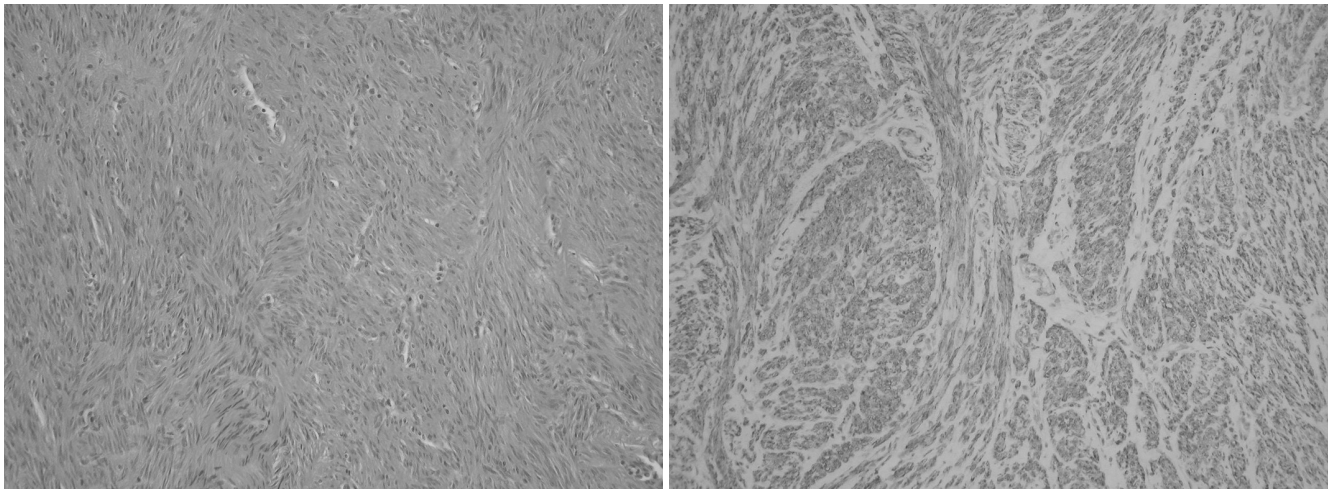
A B

**Fig. 1. A, B.** The contrast enhanced CT scan shows a lobulated, homogeneously enhancing, soft tissue mass in the right S1 paravertebral region, the pelvic cavity and within the spinal canal together with vertebral body erosion (arrows). This lesion is displacing the right psoas muscle (★) anterolaterally.



A B C

**Fig. 2. A.** The sagittal T1-weighted MR image demonstrates a soft tissue mass in the spinal canal from the L5 to S2 levels and in the prevertebral space (arrows).
**B.** The sagittal T2-weighted MR images shows the lesion has nearly isosignal intensity to the adjacent muscles.
**C.** The contrast-enhanced axial T1 image shows the extradural mass exiting the spinal canal through the bilateral neural foramina at the level of L5, and so creating a large bilobulated paraspinal component along the medial and lateral sides of the right psoas (★) muscle and displacing it anterolaterally (arrows).



A B

**Fig. 3.** Photomicrograph from the high power magnification microscopy.
**A.** The tumor shows intersecting short fascicles of spindle cells. (hematoxylin-eosin stain, ×200)
**B.** The spindle cells reveal cytoplasmic positivity upon immunohistochemical staining for smooth muscle actin. (Labelled streptoavidin biotin, ×200)

The final pathologic diagnosis was a leiomyoma without signs of malignant degeneration. Upon histologic examination, the lesions showed intersecting short fascicles of acidophilic spindle cells without any significant cellular pleomorphism or mitotic activity. No malignant features were noted (Fig. 3A). Immunohistochemical evaluation revealed a positive reaction of the tumor cells to the smooth muscle marker (smooth muscle actin) (Fig. 3B).

## Discussion

Leiomyomas are benign tumors that consist of well-differentiated smooth muscle tissue and vascular collagenous tissue, and they mainly occur in the uterus. The other tumor locations include the gastrointestinal tract, mainly in the lower third part of the esophagus, the sinonasal tract or larynx, lung, trachea, bladder, liver and adrenal gland. The tunica media and smooth muscle of the skin are also known sites for leiomyomas (7). In addition, there have been reports of primary intracranial leiomyomas and primary spinal extradural leiomyomas in male pediatric patients and adult patients with AIDS (1 - 3).

Uterine leiomyomas are on rare occasion associated with extrauterine benign smooth muscle tumors: this curious condition is referred to as "benign metastasizing leiomyoma", which is a relatively well-known phenomenon (4 - 8). Although most commonly seen in the lungs (4), other sites of metastatic involvement include the lymph nodes, peritoneum, retroperitoneal structures, spine and the base of the skull (4 - 8).

Several hypotheses have been proposed to explain the pathogenesis of this poorly understood entity. Some investigators have classified this tumor as a low grade leiomyosarcoma with malignant potential (9). Others have proposed a mechanism of implantation and proliferation of benign smooth muscle cells by an intravenous route or by mechanical means (9). Still others have postulated that this is the result of a systemic leiomyomatosis with multifocal, but independent smooth muscle proliferation (9).

We can speculate that there was tumor cell migration by an intravenous route: the most plausible explanation in this case is from the pelvic vein to the spinal epidural vein The mechanism of hematogenous spread is subendothelial vascular involvement of the uterine leiomyoma, which possibly represents early vascular invasion via the formation of a tumor embolus (10).

As far as we could establish, our patient had no evidence of metastatic disease in other organs, including the lung. Moreover, a case of a benign metastasizing leiomyoma in a spinal extradural space without lung parenchymal involvement has been reported by Hekster et al (7).

The diagnosis upon imaging was initially mistaken as a neurofibroma because of its location and its characteristic shape. Yet after retrospectively reviewing the MR images, we found that the large mass exhibited lower T2 signal intensity than that of a neurofibroma, and it

showed rather strong homogeneous enhancement on the gadolinium enhanced T1 weighted images.

Complete resection of these lesions is usually possible and this can provide substantial improvement of the patient's functional status. Because of this potential for successful treatment, this disease entity should be kept in mind when diagnosing a paraspinal lesion that exhibits unusual imaging features.

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# 신경섬유종으로 오인된 척수에 생긴 전이성 양성 평활근종: 증례 보고[1]


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