Upsala J Med Sci 108: 213–220, 2003

# Leiomyosarcoma of the Sacral Bone in a Patient with a Past History of Resection of Uterine Leiomyoma

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## ABSTRACT

We report here a case of leiomyosarcoma in the sacrum with a differential diagnosis of metastasizing leimyoma of uterus, since the patient had a past history of resection of uterine leimyoma 19 years ago. The sacral tumor was an osteolytic lesion, 8×6 cm in size on radiological examination. Microscopically, tumor cells consisted of spindle shaped cells with moderate cellular atypia. The tumor cells invaded into the surrounding muscle tissue. Immunohistochemistry revealed that the tumor cells were positive for α-SMA and vimentin, and they were enclosed by type IV collagen, suggesting the presence of the basement membrane. The labeling index of Ki-67 in the tumor cells was 25 %. Re-examination of leiomyomas of uterus resected 19 years ago showed that they were typical leiomyomas, showing well-circumscribed tumors, composed of well-differentiated smooth muscle cells without nuclear atypia. The presence of radiological and pathological findings of malignancy of the sacral tumor excluded the possibility of metastasizing leiomyoma, suggesting that the sacral tumor was another primary tumor.

## INTRODUCTION

Leiomyosarcoma is a malignant tumor, usually arising in the soft tissues. When such tumor is detected in bone tissue, we should first examine whether the bone lesion is metastatic or original. We experienced a rare case of leiomyosarcoma arising in the sacrum. The patient had a past history of the resection of uterine leiomyoma 19 years ago. Uterine leiomyoma on rare occasion metastasizes to the lung despite its benign pathological findings. They are designated as “metastasizing

Received 26 August 2003
Accepted 26 September 2003

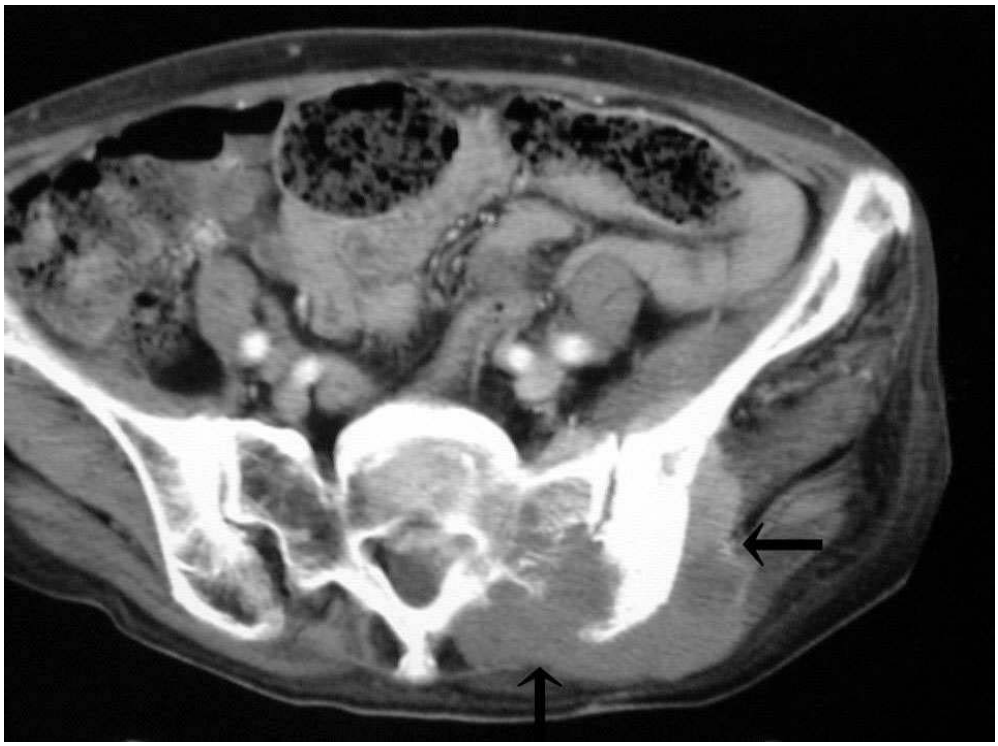
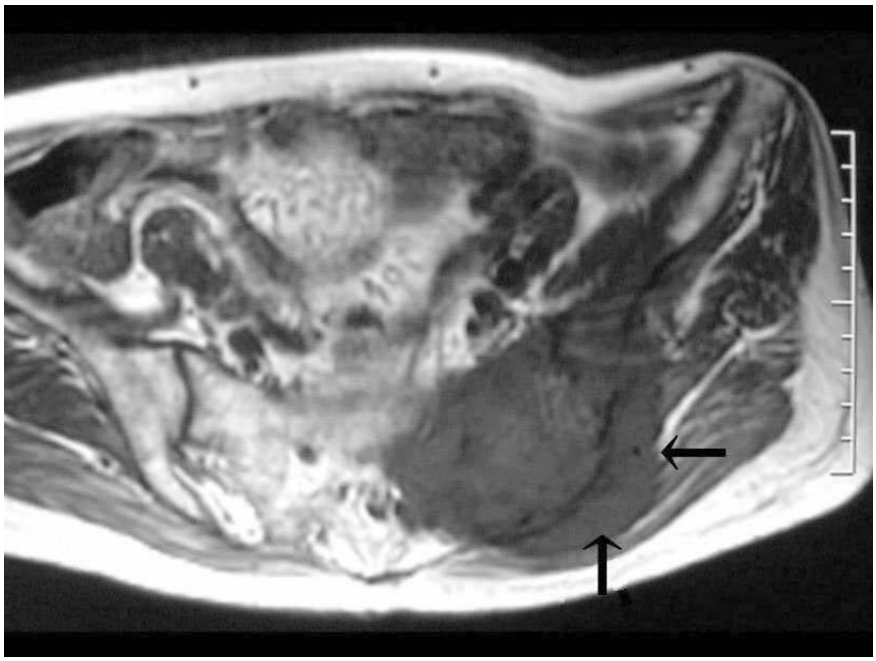


*Fig. 1.* Enhanced CT image. Bone destruction of the sacrum and left ilium is detected, enhanced chiefly in the peripheral areas.

leiomyoma" (2, 5, 8). We report the present case with careful consideration whether the sacral tumor was the second original tumor or metastasizing leiomyoma from the uterine leiomyoma.

## CASE REPORT

67-year-old woman was admitted with a chief complaint of left buttock pain. On X-ray radiograph, a bone-destructive lesion was detected in the sacrum and left ilium. On CT image, the tumor showed an osteolytic lesion, 8×6 cm in size, and was enhanced chiefly in its peripheral areas (Fig. 1). MRI reveals that the tumor was of low intensity compared with muscle tissue on T1-weighted image (Fig. 2a). The tumor showed an enhancement in the peripheral areas after gadolinium injection (Fig. 2b). The tumor showed moderate central hyperintensity on T2-weighted image. Tumor invasion into the internal pelvic organs was not detected on images. The needle biopsy was done from the central part of the tumor, and the open biopsy was done one month later. The pathological findings were almost the same. The tumor was composed of proliferation of atypical spindle-shaped cells in a fascicular pattern (Fig. 3 a). Mitoses were observed with a mitotic index less than 1 per 10



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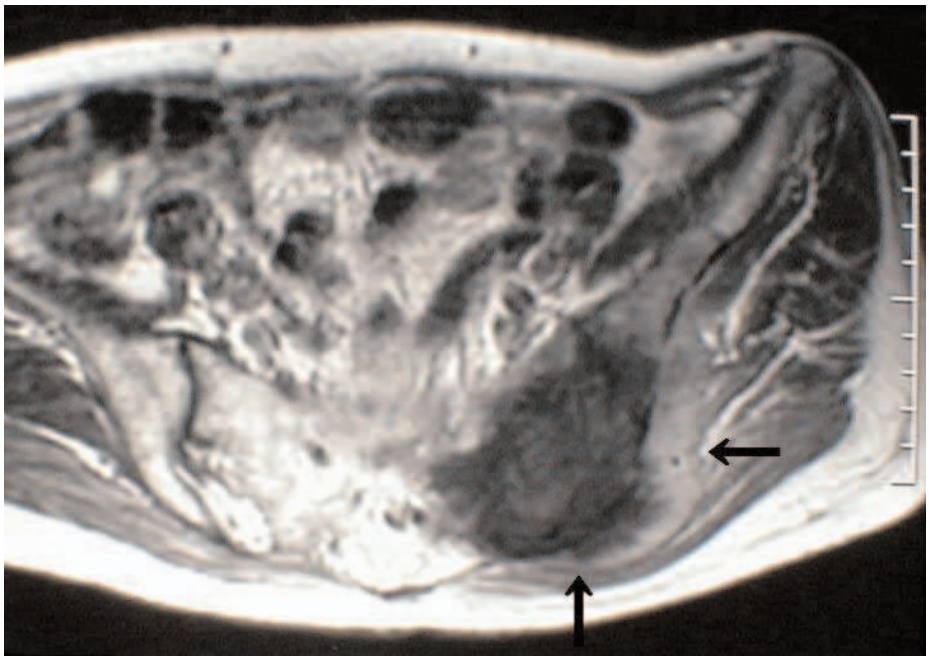

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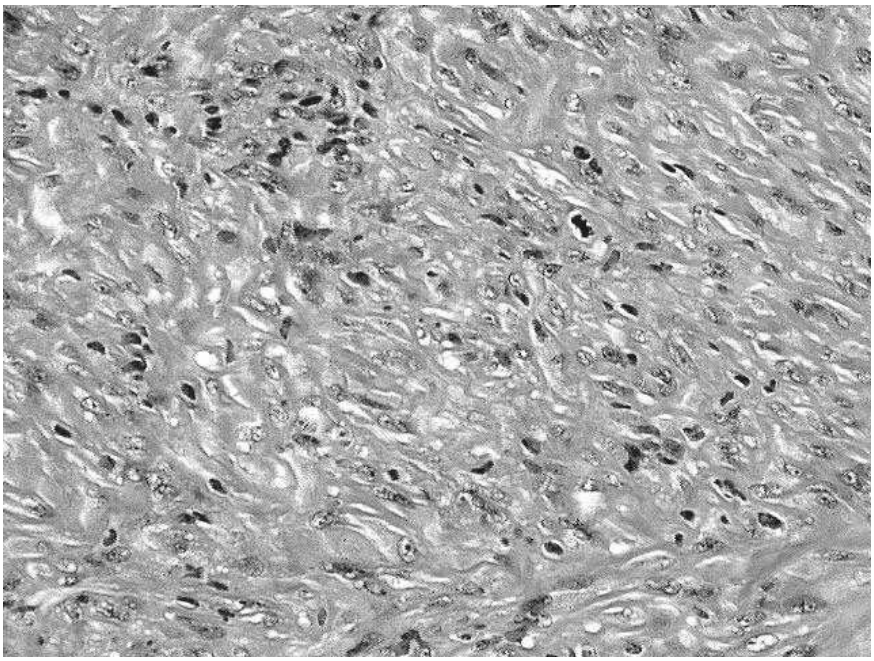
*Fig. 2.* Three MRI images showing low intensity in tumor compared to muscle on T1-weighted image (a), peripheral enhancement after gadolinium injection (b).



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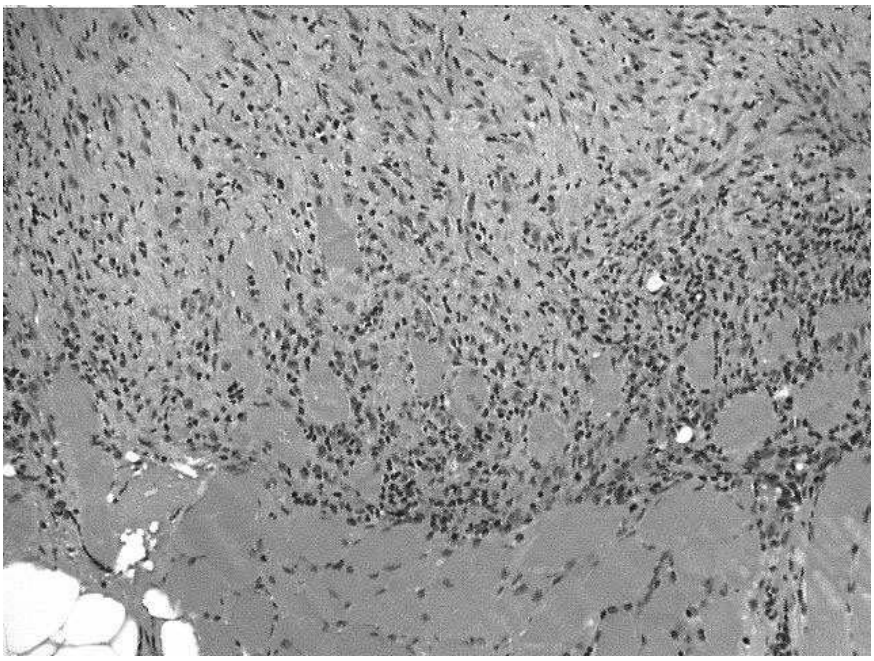

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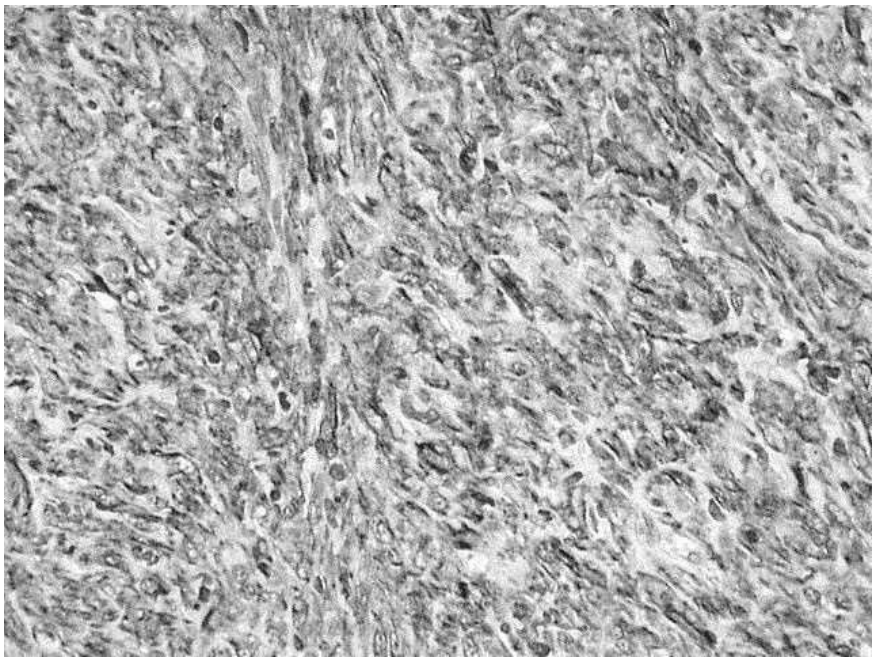
*Fig. 3.* HE stain of the sacral tumor tissue. Spindle cells irregularly proliferate forming bundles (a). The tumor cells invade into the surrounding muscle tissue (b).



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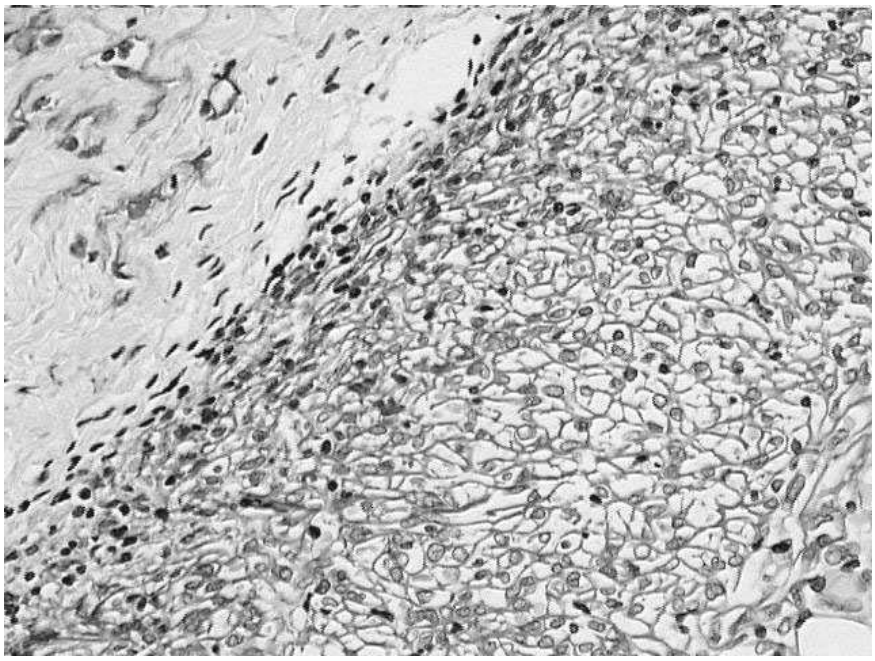

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*Fig. 4.* Immunohistochemistry of the sacral tumor tissue. α-smooth muscle actin is positive in the cytoplasm (a). Almost all tumor cells are enclosed by type Ⅳ collagen (b).

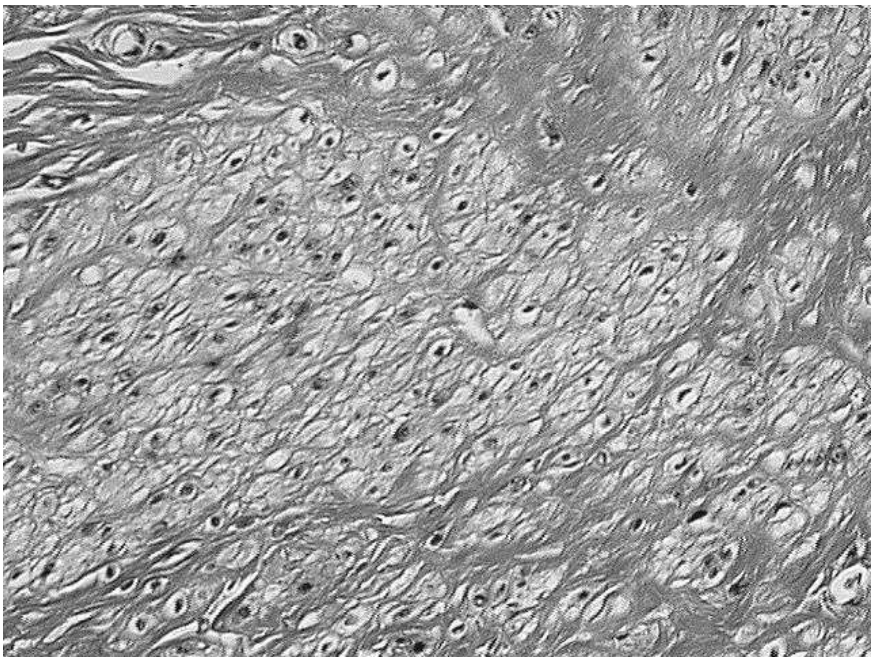

*Fig. 5.* HE stain of uterine leiomyoma showing benign features.

high power fields. The coagulation necrosis was not seen. The tumor cells invaded into the surrounding muscle tissue (Fi. 3 b). Therefore, low-grade spindle cell sarcoma was suspected.

Immunohistochemically, the tumor cells stained positively for vimentin, α-smooth muscle actin (αSMA) (Fig. 4 a), and muscle specific antigen (clone HHF-35). Desmin was positive in a part of tumor cells. These immunohistochemical findings suggested either smooth muscle origin or active fibroblast (myofibroblast)-origin. Immunohistochemistry for type IV collagen can be useful for the differential diagnosis. Almost all the tumor cells were enclosed by type IV collagen (Fig. 4 b). This indicated the presence of basement membrane, supporting the smooth muscle-origin. Ki-67 was positive in 25 % of the tumor cells. The tumor cells did not stain for S-100 protein, cytokeratin, CD34, or c-kit. The sacral tumor was diagnosed as leiomyosarcoma. Metastatic tumors in other sites or other primary tumors were not detected by the whole body examinations including chest-abdominal CT scan, technetium & gadolinium scintigraphy, and gastrointestinal endoscopic examination.

The patient had a past history of resection of leimyoma of uterus, performed 19 years ago. The uterine tumors were re-examined. On gross examination, approximately ten clearly margined whitish tumors were detected. Microscopic observation revealed proliferation of spindle-shaped cells. Neither cellular atypia nor mitosis was found (Fig. 5). Immunohistochemically, the tumor cells stained for vimentin, a-

SMA, and desmin, and they were surrounded by type Ⅳ collagen. Ki-67 was a positive in 0.4% of the tumor cells. Thus we corroborated the original diagnosis of uterine leiomyoma, confirming the histological differences from the sacral tumor. The patient received radiation therapy after the resection of sacral tumor. The state of the whole body deteriorated afterwards, and she died of cachexia two years later. Autopsy was not permitted. Recurrences, metastases or other primary tumors were not detected through the whole clinical course.

Leiomyosarcoma is a rare bone tumor, comprising 0.1% of all primary bone tumors (6). Miura reviewed 136 cases of leiomyosarcoma of bone reported from 1944 to 1999 (7). In 108 of 136 cases, the bone origination was confirmed by radiographical observation. The average year was 49, ranging from 9 to 87 years old. No sex predominance was observed (56 men, 52 female). Femurs were most frequently affected (39%), followed by tibias (23%), pelvis (12%), humerus (7%), mandibles (7%), and other sites (11%). It is well known that uterine leiomyoma can metastasize to the lung, even though it is seemingly benign (1, 3, 4). Such "benign metastasizing leiomyomas" were reported in 54 cases from 1973 to 1999. Among them, there is a case of low grade leiomyosarcoma arising 15 years after hysterectomy (4). It remains unsolved whether such cases should be included in metastasizing leiomyoma or not. In the present case, the metastasis was not seen in the lungs and the histological features of tumors in the uterus and sacrum were different. Therefore, we diagnosed that the uterine tumors was leiomyomas and the sacral tumor was an independent leiomyosarcoma.

## ACKNOWLEDGEMENTS

We are grateful to Dr. Fumiaki Tezuka, Dr. Masami Hosaka, Dr. Takashi Suzuki, Dr. Takuya Moriya for their valuable suggestions for pathological examinations.

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