Metastatic skeletal leiomyomatosis
(leiomyomatosis ossea)

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Abstract We present a unique case of metastatic leiomyomatosis to the skeleton. The very extensive involvement of the axial and peripheral skeleton with “ring” lesions and associated cyclical premenstrual pain eventually led to the correct diagnosis and total relief with hormonal therapy.

Keywords Uterine leiomyomatosis · Cyclical pain · Skeletal “ring lesions” · Radiographs · MRI

Introduction

The involvement of the lungs by metastases arising from benign leiomyomas of the uterus is well known although uncommon. [1] Spread of these metastases to the skeleton is exceedingly rare and has only recently been reported [2] The case reported here is unique because of the widespread lesions involving the axial and peripheral skeleton with the sparing of the lungs. The association of skeletal symptoms with cyclical premenstrual pain and complete relief from hormonal therapy is documented and discussed.

Case report

A 39-year-old Brazilian woman presented with severe lumbar pain in 1990 (at the age of 30 years). She was then in the latter part of her second pregnancy. The pain radiated down both legs to the knees but predominantly on the left side. It was accompanied by paresthesiae and weakness of the left leg while the pain lasted. She was treated as having sciatica. Following parturition in February 1991 the pain disappeared completely but returned with renewed strength in November 1991. She then underwent complete investigation. She had radiographs of her spine, pelvis and upper femora (Figs. 1, 2), scintigraphy (not shown), CT (not shown) and MR imaging of her spine (Fig. 3).

The radiographs were very striking, showing multiple areas of lucency with sclerotic rims in both iliac wings and upper femora. The vertebral also showed a multiplicity of lesions. These varied in size from 1 to 2 cm. The L2 vertebra showed erosion of the posterior cortex with a multilocular lesion extending into the left half of the vertebra. This was the largest single lesion.

The radiological diagnosis at this time was thought to be Langerhans cell granuloma (eosinophilic granuloma) and the patient was advised to have radiation therapy to her L2 vertebra. This was delayed until she had a biopsy of one of the ring lesions in the neck of her left femur. This was an open biopsy performed in April 1992 and the abundant material was interpreted as non-ossifying fibroma of bone. Despite this diagnosis, the L2 vertebra was irradiated. The dose was 4000 cGy. There was no improvement at all in her severe back pain.

It was about this time that the patient noted for the first time a cycli-
cal pattern to her pain, which appeared to be related to her menstrual cycle. The pain started in the premenstrual phase (about 10–12 days), being relatively mild in the beginning; it became progressively more intense and severe for 5–6 days and then gradually diminished until 2–days before her menstrual period, when it disappeared completely. The patient remaining completely free of pain and asymptomatic for 18 days. The cycle was then repeated each month. The pain was so severe that she frequently required morphine to control it.

In October 1992 it was decided to biopsy the L2 vertebra. This was a closed biopsy with a Craig needle. The material, which was sparse, was interpreted as a lesion with fusiform cells, probably benign. A repeat biopsy was not performed. The patient in desperation saw many physicians between 1992 and 1998 and also paramedical practitioners. However, it was an acupuncturist, impressed by the constant relationship of the pain to her menstrual cycle, who referred her to an endocrinologist. She in turn referred the patient to a gynecologist because of her previous history of leiomyomas of the uterus. She found a bulky uterus and following preoperative evaluation the patient underwent a total hysterectomy and right adnexitomy.

Histologically the uterus showed multiple benign leiomyomas, hyper-

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**Fig. 1** Frontal radiograph of the pelvis (1992). There are multiple lucent lesions in both hemipelves with sclerotic rims. Some are relatively low in density and poorly defined. The lesion in the left acetabulum is not well seen. Patchy sclerosis present in the left supra-acetabular region. There are prominent lesions in the left upper femur

**Fig. 2** Frontal radiograph of the pelvis (1999). The “ring” lesions are very much better defined and much more profuse in all the bones of the pelvis. The left acetabular lesion has a fine rim of density surrounding quite a large lesion. Note there is much more sclerosis in the lesion above the acetabulum

**Fig. 3** MR images of the lumbar spine (1998). A Sagittal SE T1-weighted image with gadolinium (TR/TE 400/16). A bulky mass in L2 is compressing the dura with associated increased signal. Note the multiple lesions in the other vertebrae. B Axial SE T1-weighted image (TR/TE 745/20) with gadolinium. Note the erosion of the vertebra with a bulky mass and increased signal markedly deforming the dura sac