

The rare presentation of sinus tarsi syndrome secondary to metastasis in a patient with endometrial carcinoma

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Metastasis to bone or synovial tissue in the distal extremity is a rare event for endometrial carcinoma. This case report involves a 76-year-old female with stage II endometrial cancer who developed persistent ankle pain after a transabdominal hysterectomy and bilateral salpingo-oophorectomy. Plain films were initially negative. An MRI several weeks later demonstrated an abnormality in the synovial tissue of the sinus tarsi and in the distal tibia. A bone scan revealed increased activity on both sides of the ankle joint. Soft-tissue biopsy of the sinus tarsi was positive for endometrial adenocarcinoma. This is the first reported case of sinus tarsi syndrome, a chronic pain condition in the sinus tarsi of the hindfoot, secondary to metastatic endometrial carcinoma.

Introduction

Endometrial carcinoma is the fourth most common cancer diagnosed in American women, following cancers of the breast, lung, and colon (1). Ninety percent of the cases present with uterine bleeding and occur in women over age 50 years, with a median age of 61 years (2). Seventy-two percent of endometrial cancers present at stage I, 12% at stage II, 13% at stage III, and 3% at stage IV (2). Whereas 5-year survival for stage I disease may approach 90%, stage IV survival ranges from 10% to 15% (3).

Endometrial carcinoma usually spreads either by direct invasion or via lymphatics to abdominopelvic sites. However, hematogenous dissemination to distant sites such as

the lung, liver, or bone may occur (4). A recent review of 29 case reports of bone metastases from endometrial carcinoma (5) found that lesions often involved appendicular skeletal sites, particularly the calcaneus (21%). However, the largest study of endometrial bone metastases, based on an autopsy series ($n=67$), found that 25% of the patients had bone metastases, with vertebrae (76%) being the most frequently involved site (6). Metastatic spread to synovial tissue in a joint space has not been previously reported for endometrial carcinoma (7).

The patient in our case presented with stage II endometrial cancer and postoperatively developed persistent left-ankle joint pain. Joint pain has a wide range of differential diagnoses, including inflammatory, traumatic, infectious, and (less commonly) malignant causes. Arthritis related to metastatic disease is often secondary to lung cancer and typically affects the knee joint (8). We report a rare case of sinus tarsi syndrome (a chronic pain condition involving the sinus tarsi, or talocalcaneus sulcus, of the hindfoot) secondary to the metastatic spread of endometrial carcinoma.

Case report

A 76-year-old nulligravid woman presented with postmenopausal bleeding and pelvic cramping. Physical exam was otherwise unremarkable. Dilation and curettage revealed grade 2/3 endometrioid adenocarcinoma with squamous areas. Higher-grade endometrioid tumors repre-

Citation: Pieters RS, Galvin J. The rare presentation of sinus tarsi syndrome secondary to metastasis in a patient with endometrial carcinoma. *Radiology Case Reports*. [Online] 2011;6:414.

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Competing Interests: The authors have declared that no competing interests exist.

DOI: 10.2484/rcr.v6i2.414

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Figure 1. 75-year-old female with sinus tarsi and endometrial carcinoma. T1 sagittal image demonstrating tumor in sinus tarsi and focal tumor in distal tibia.

sent a less well-differentiated tumor type associated with a worse prognosis (9). Abdominopelvic CT, mammography, and a preoperative chest radiograph were negative. She underwent transabdominal hysterectomy, bilateral salpingo-oophorectomy, node dissection, and washings within three weeks. Washings, nodes, and adenexa were negative. The tumor involved the inner half of the myometrium and endocervical stroma, with lymphovascular invasion. The patient was referred for postoperative radiotherapy for FIGO pathologic stage IIB, T2bN0MX, endometrial carcinoma. She received postoperative pelvic radiotherapy with boosts to the pelvic sidewalls and endovaginal brachytherapy to boost the vaginal wall.

Surgical recovery was uneventful except for the onset of pain in the left ankle with weight-bearing. The pain progressed over 6 weeks until radiation therapy was started. Plain-film radiographs (not available) demonstrated only some soft-tissue swelling; the bony structures and joint spaces were unremarkable. When the pain and swelling progressed, an MRI was obtained about 10 weeks postoperatively.

MRI (Figs. 1-5) demonstrated an abnormal process centered in the sinus tarsi with involvement of the anterior process of the calcaneus. A smaller lesion was seen in the posterior aspect of the distal tibial metaphysis, associated with possible cortical breakthrough and periosteal elevation. Sagittal (Figs. 2A and 2B) and axial (Figs. 3A and 3B) views demonstrate cortical disruption of the anterior process of the calcaneus in association with the mass in the sinus tarsi. There is adjacent reactive edema. Fig. 4 illustrates

the distal tibial lesion with surrounding reactive edema. Fig. 5 demonstrates the involvement of the anterior calcaneal process.

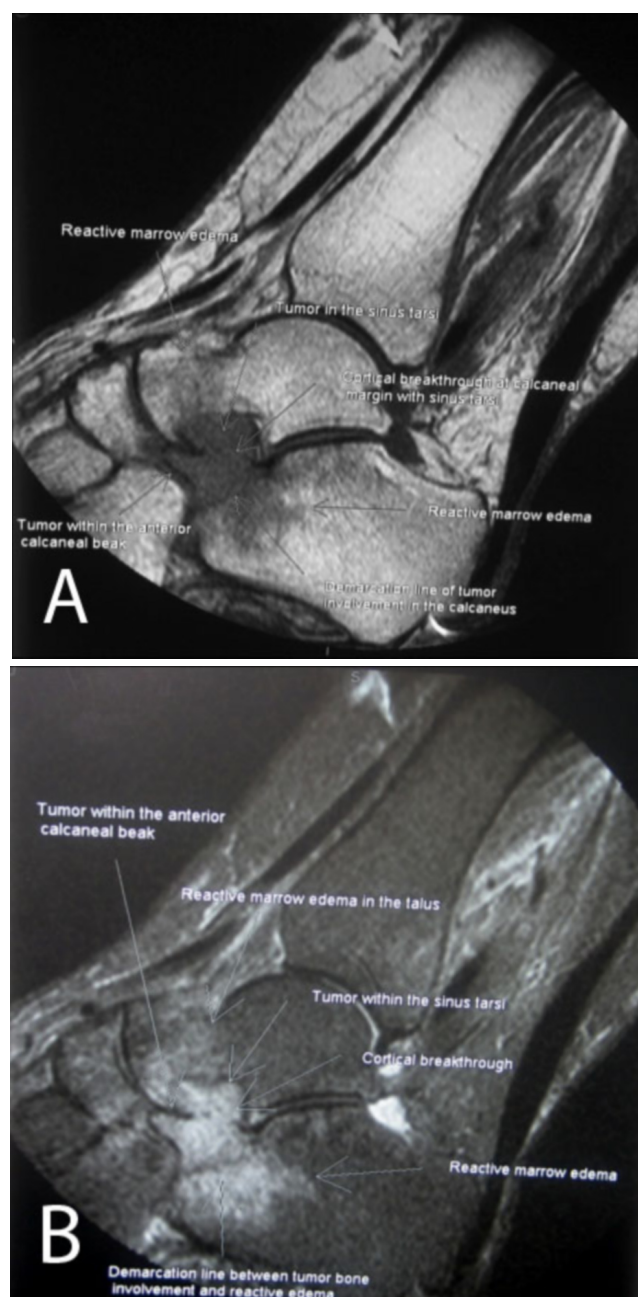


Figure 2. 75-year-old female with sinus tarsi and endometrial carcinoma. A: T1 sagittal image demonstrating tumor in sinus tarsi and adjacent bone with reactive edema; B: STIR sagittal image.

The initial bone scan (not available) was positive in the ankle, with increased activity on both sides of the left ankle joint. Plain films at that time (not available) demonstrated lucency and expansion of the anterior superior process of

Sinus tarsi syndrome secondary to metastasis in a patient with endometrial carcinoma



Figure 3. 75-year-old female with sinus tarsi and endometrial carcinoma. A: T1 axial image at sinus tarsi level. B: STIR axial image at sinus tarsi level.

the calcaneus. On physical examination, the foot was erythematous, warm, exquisitely tender, and swollen anteriorly, but there was no fever. Overall, the pattern of involvement was thought unusual for metastatic disease, favoring primary malignancy, acute infectious or inflammatory process, or a benign proliferative or deposition disease. The diagnostic radiologist reported that the bone scan findings corresponded to previous MRI images and were most consistent with a process arising from the synovium in the sinus tarsi and extending into bone. The epicenter was thought to be extraosseous.

The patient was referred to orthopedic oncology, with a presumptive diagnosis of a sarcoma, a second primary.

Soft-tissue biopsy demonstrated adenocarcinoma, consistent with the previous endometrial malignancy. Restaging CT scans of chest, abdomen, and pelvis were again negative.

Palliative radiotherapy was initiated, and the pain responded rapidly. The metastatic lesions in the sinus tarsi, calcaneus, and tibia were treated as an isolated metastasis, as all sites could be encompassed in a single radiotherapy portal. The patient required low-dose narcotic analgesia for foot pain. She began Tamoxifen, 20 mg daily, when radiation treatment was completed.



Figure 4. 75-year-old female with sinus tarsi and endometrial carcinoma. STIR coronal image demonstrating tumor in distal tibia.

Repeat bone scans 6 and 12 months after the onset of foot pain revealed continued increased activity in the left ankle, though diminished from the initial scan. Restaging abdominal and pelvic CT scans remained negative.

Approximately two years after her initial presentation, the patient developed pain and swelling in the left thigh. An MRI (Figs. 6-7) revealed a 6-cm marginated mass in soft tissue along the posterior aspect of the mid-left femur, possibly representing a second area of synovial involvement (along a tendon sheath). Restaging CT scans were again otherwise negative. As it was the only involved site, she was referred for consideration of resection. Diagnostic biopsy confirmed a metastasis consistent with endometrial carcinoma. Orthopedic oncology declined to resect this lesion. She was then referred for palliative radiotherapy, but the

Sinus tarsi syndrome secondary to metastasis in a patient with endometrial carcinoma

pain failed to resolve after this treatment. Repeat staging 2 months later revealed that the patient was free of disease except for the known deposit of metastatic disease of the left thigh. The patient continued on narcotic analgesia for chronic pain and was placed on low-dose doxorubicin. Six months later, the patient developed bone, adrenal, and liver metastases and a pelvic nodal mass, which did not respond to chemotherapy. She went on to expire three years after primary diagnosis.



Figure 5. 75-year-old female with sinus tarsi and endometrial carcinoma. STIR coronal image demonstrating tumor billowing out of sinus tarsi.

Discussion

The sinus tarsi is the joint space between the calcaneus and talus that contains ligamentous and neurovascular structures. The syndrome, marked by pain, swelling, and subtalar joint instability, is usually related to trauma (10). A review of bone metastases associated with endometrial cancer revealed only two cases involving the talocalcaneal region without other distant metastases (5).

In the case presented, tumor clearly involved the sinus tarsi. Whether the tumor originated in the sinus tarsi and subsequently invaded bone or whether the tumor extended out of the anterior calcaneus into the sinus tarsi is unclear. Tumor was also clearly identified within the distal tibia. The subsequent thigh lesion, with involvement along the tendon sheaths of the posterior femur (without bone in-

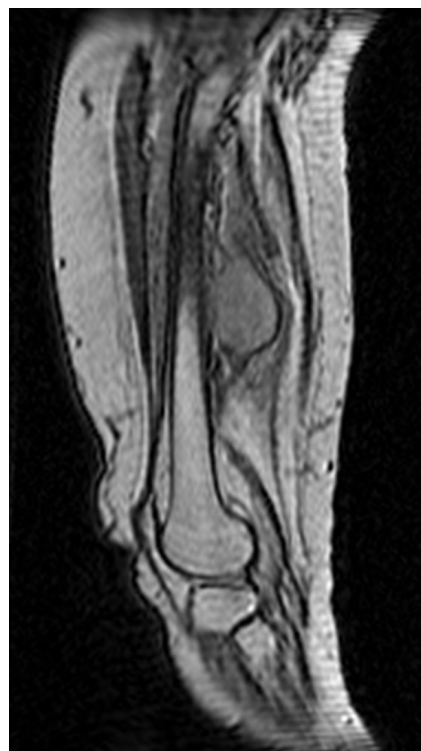


Figure 6. 75-year-old female with sinus tarsi and endometrial carcinoma. Localizing sequence sagittal image demonstrating a thigh metastasis.

volvement), suggests that the tumor may have spread directly to both synovium and bone in this patient. Synovial metastases, though rare, usually occur with lung cancer and have not been previously reported for endometrial carcinoma (7). Currall et al. (7) reviewed reports of synovial metastases and proposed that the likely mechanism of synovial metastases is via direct

spread from a periarticular bony metastasis. They found that the majority of 39 cases of synovial metastases had radiologic changes and/or bone-scan abnormalities, whereas both tests were negative for only two cases. In our case, initial plain films were negative, but the bone scan 6 weeks later was positive.

Metastatic disease is often associated with a poor prognosis. Median survival time after bone metastases associated with endometrial cancer is about 16 months (5). Survival after discovery of synovial metastases usually averages less than five months (7). Oligometastatic disease, defined as disease to a single or limited number of organs (11), may carry a more favorable prognosis. In our case, the patient was perceived as having an isolated metastasis, even though multiple bones were involved. A decision was made to proceed with endovaginal brachytherapy, and pelvic control was maintained until the end stage of her disease.

In summary, metastatic disease should be suspected in any patient with a history of cancer who presents with localized joint pain, particularly if it is unresponsive to analgesia. MRI cannot differentiate benign from malignant lesions, so tissue biopsy is necessary for diagnosis (10). Finally, an isolated metastasis or oligometastatic disease may have a more favorable prognosis, and radical treatment of such lesions warrants further study.

Sinus tarsi syndrome secondary to metastasis in a patient with endometrial carcinoma

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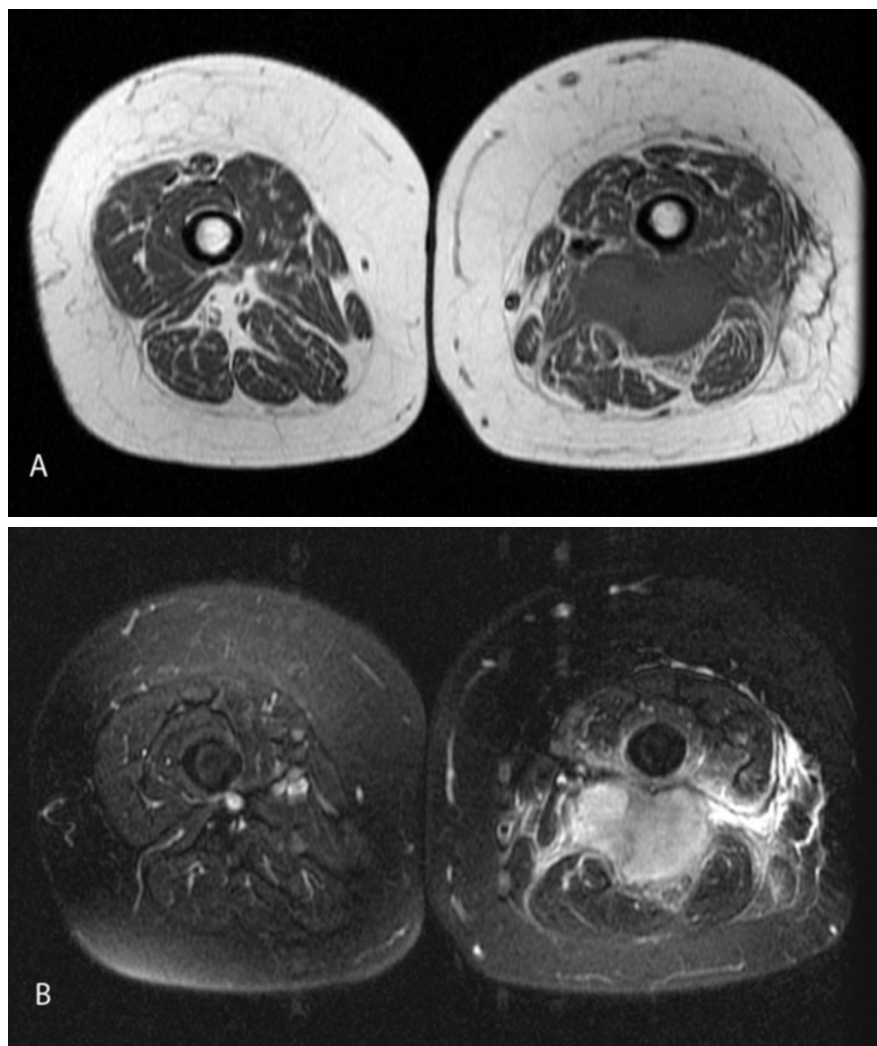


Figure 7. 75-year-old female with sinus tarsi and endometrial carcinoma. A: T1 axial image demonstrating a thigh metastasis; B: T2 axial fat-saturated sequence demonstrating a large soft-tissue mass without bone involvement.