

Radiation-induced periosteal osteosarcoma

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Abstract

Radiation-induced sarcomas are a well-recognized late complication of radiation therapy. We present a rare case of a periosteal osteosarcoma of the femur developing 10 years after radiation treatment for a soft tissue sarcoma to highlight the clinical and radiological features of this rare complication of therapy and to describe its management.

Keywords

Femur; periosteal osteosarcoma; radiation sarcoma; MR imaging.

Introduction

Radiation-induced sarcomas of bone are rare but well recognized, accounting for approximately 1.5% of all bone sarcomas^[1]. Estimates of the incidence range from 0.03% of patients who receive radiation treatment to 0.2% of those who survive 5 years^[2]. The latent period between radiation treatment and presentation of the sarcoma ranges from 4 to 55 years (mean = 11 years)^[1,3,4]. Over 90% of cases are high-grade intramedullary osteosarcoma or spindle cell sarcoma (fibrosarcoma and malignant fibrous histiocytoma)^[5]. The purpose of this report is to present the first fully documented example in the English language literature of a periosteal osteosarcoma occurring 10 years after radiation treatment for a soft tissue sarcoma and to highlight the diagnostic and management features.

Case report

A 52-year-old female presented with a 2 month history of pain and swelling in the lateral aspect of the thigh. A magnetic resonance (MR) imaging scan revealed the typical appearance of a soft tissue sarcoma arising in the vastus lateralis muscle, measuring 20 cm in length. Further staging studies showed the lesion to be solitary with no evidence of metastatic disease. Needle biopsy confirmed a high-grade soft tissue malignant fibrous histiocytoma. The tumour was excised with a wide margin, including the underlying vastus intermedius but leaving the periosteum of the femur intact and the patient received postoperative radiotherapy. In total 66 Gy in 33 fractions over 7 weeks were administered over the lower 25 cm of the lateral half of the thigh. Follow-up was unremarkable for 5 years. A year later the patient complained of some discomfort in the thigh and radiographs revealed a very thin horizontal crack fracture through the lateral cortex of



Fig. 1. Anteroposterior radiograph of the femur showing dense amorphous periosteal new bone formation along the lateral aspect of the femoral diaphysis.

the mid-femoral diaphysis. This was considered to be a stress fracture possibly secondary to early radiation damage to the cortex and her symptoms settled with conservative management.

Four years later, now 10 years after treatment of the primary tumour and radiotherapy, the patient re-presented with increasing discomfort over the lower thigh. Radiographs showed irregular, amorphous periosteal new bone formation arising over the lateral aspect of the distal femoral diaphysis (Fig. 1). Whole-body scintigraphy showed increased activity over the lesion and a tiny focus more proximally corresponding to the previously identified stress fracture (Fig. 2). Computed tomography (CT) showed generalized thickening and lucencies within the distal femoral diaphyseal cortex indicative of radiation damage with an area of irregular periosteal new bone formation; MR imaging confirmed a surface lesion of bone with a deeper mineralized component and a peripheral enhancing soft tissue component (Fig. 3). The imaging diagnosis was presumed to be a surface osteosarcoma. A needle biopsy confirmed a periosteal osteosarcoma of intermediate grade with prominent chondroblastic elements. Wide excision of the tumour was performed with endoprosthetic replacement of the distal 25 cm of the femur. Postoperatively the patient received three courses of chemotherapy with adriamycin and cisplatin.

Macroscopic examination of the resected specimen showed a fusiform mass surrounding the distal femur (Fig. 4). At sectioning the lesion was grayish-white with prominent cartilagenous areas. The periosteum was stretched over its surface. The underlying cortex and medulla were

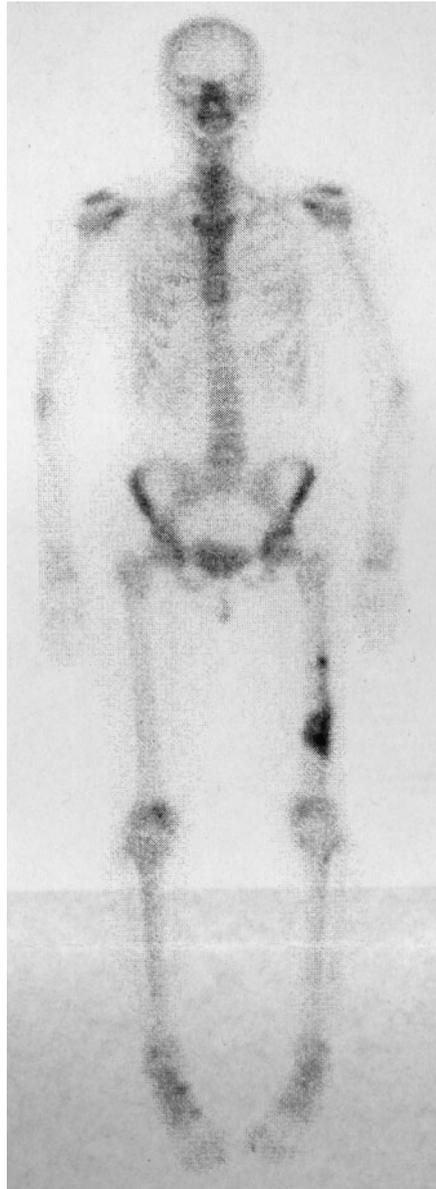


Fig. 2. Anterior projection of whole-body bone scintigraphy showing increased activity over the lateral aspect of the left femoral diaphysis. The small focus of increased activity located proximally represents the previously identified stress fracture.

uninvolved. Histologically, the tumour had the characteristic features of a periosteal osteosarcoma with lobules of atypical cartilage that were separated by primitive atypical mesenchymal cells (Fig. 5). Areas of osteoid production and foci of calcification were noted in the central parts of the lesion. The patient remains disease free 4 years later with good function in her limb.

Discussion

Malignant transformation of bone following radiotherapy was first documented by Beck in 1922^[6]. Reports of many large series of radiation-induced sarcomas have since been published comprising 700 cases in total. In none of these series is there a description of a surface osteosarcoma^[1,7,8]. Similarly in series of periosteal and high-grade surface osteosarcomas^[9-11] and parosteal osteosarcomas^[12,13], there was no reported association with radiation treatment. In the author's own unit with experience of treating 42 radiation-induced bone sarcomas and 31 periosteal osteosarcomas, this current case was the only one common to both groups^[3,11]. One outcome study of 23 radiation-induced osteosarcomas, two surface osteosarcomas are mentioned

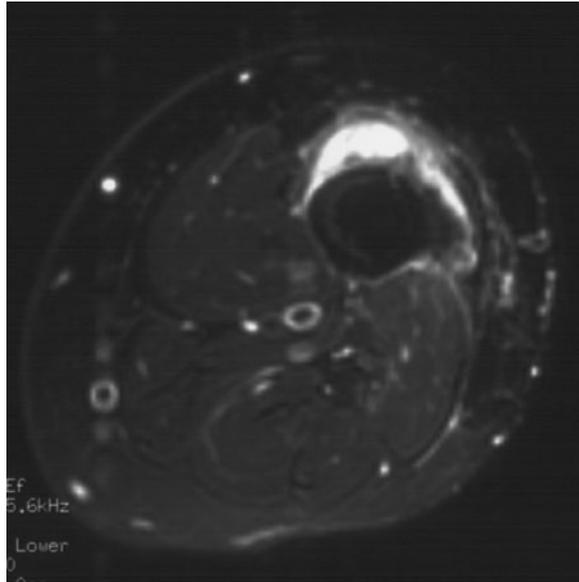


Fig. 3. MR imaging of the femur: axial T1-weighted fat suppressed contrast-enhanced image showing the persistent low signal intensity of the mineralization and marked enhancement of the peripheral soft tissue component of the lesion. The medulla was uninvolved on all sequences.

briefly; one was a high-grade surface osteosarcoma and the other was a periosteal osteosarcoma^[14]. A literature search revealed two further examples of radiation-induced surface osteosarcoma: a parosteal osteosarcoma arising on the femur in a 16-year-old, 14 years after radiation treatment for an eosinophilic granuloma^[15] and a further parosteal osteosarcoma of the mastoid bone 5 years after radiation therapy for a nasopharyngeal carcinoma^[16]. Therefore, radiation-induced surface osteosarcoma is rare. The current case is the first fully documented example of a periosteal osteosarcoma in the English language literature. Why the periosteal tissues remain resistant to the mutagenic effects of ionizing radiation compared with the underlying bone remains unknown.

In this case the tumour clearly arose under the periosteum, which had been left behind at the time of initial resection of the soft tissue sarcoma. Resecting the periosteum does increase the risk of patients developing fractures if they have radiotherapy but may, in this case, have increased the risk of second malignancy^[17].

In a patient who has previously received radiotherapy, the presence of pain accompanied by the appearance of new bone formation on a plain radiograph must be considered to be a potential indication of second malignancy and should be investigated as such.

In this particular case, the decision to resect all of the bone that had been exposed to radiotherapy (25 cm) resulted in a very large tumour resection but it was felt that this was necessary because of the potential risk of further malignancy arising in the remaining periosteum that had been exposed to radiation^[18]. Despite the known increased risks of radiotherapy in patients who receive radiation, the early results in this case are promising^[19].

The reason why individuals develop radiation-induced bone sarcomas remains a mystery and thus far no likely cause has been identified. Although genetic factors have been postulated this remains to be clarified^[20].

Teaching point

Periosteal tissue unlike the underlying bone remains resistant to the mutagenic effects of ionizing radiation. The precise cause of ionizing radiation-induced periosteal sarcoma is unknown although genetic factors have been suspected. When a patient treated with radiotherapy in the past for an osteosarcoma presents with suspicious clinical and radiological features, the possibility of second malignancy must be considered and appropriate investigations and treatment should be arranged. Surgical excision was used for complete tumour resection in this case.



Fig. 4. Sagittal cross-section of the femur showing the ossified surface lesion arising on the surface of the thickened cortex.

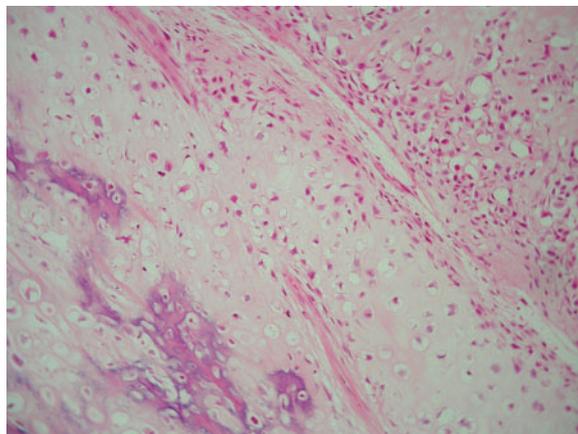


Fig. 5. Photomicrograph (H&E $\times 20$) of the tumour showing lobules of malignant cartilage with foci of chondro-osseous matrix.

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