Case Report

Rare Presentation of Radiation-induced Sarcoma Detected on F-18 FDG Positron Emission Tomography/Computed Tomography in a Treated Case of Giant Cell Tumor

Abstract

Giant cell tumors (GCTs) are benign bone lesions which are treated with curettage and bone grafting. Infrequently, GCTs show local site recurrences which are then treated with either surgical excision or radiation therapy. Radiation-induced sarcoma is rarely seen as a late complication of radiation therapy which needs to be differentiated from recurrent GCT. We report one such rare case of radiation-induced sarcoma detected on Flourine-18 fluorodeoxyglucose (18F FDG) positron emission tomography/computed tomography in a 40-year-old male who was treated with radiation therapy for recurrent GCT 9 years ago.

Keywords: *FDG positron emission tomography/computed tomography, giant cell tumor, radiation-induced sarcoma*

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Introduction

Giant cell tumors (GCTs), also known as osteoclastomas, most commonly occur in the epimetaphyseal region of femur and tibia. It can rarely (4%–10%) occur at other sites involving axial skeleton such as sacrum and vertebral bodies.[1] Although benign in nature, local site recurrence is seen in approximately 10%–20% of GCTs on the long-term follow-up which are further treated with surgical excision.[2] Due to multiple sites of involvement by the tumor, the patient is often not an ideal candidate for surgery. In such cases, other adjuvant therapies such as radiation therapy are recommended.[3] Although the benefits of radiation therapy outweigh the side effects; rarely, secondary neoplasms such as sarcomas can occur in these patients as a delayed complications on the long-term follow-up. We report one such rare case of metastatic sarcoma detected on F-18 FDG positron emission tomography/computed tomography (PET/CT) in a known case of recurrent GCT of sacrum, who was deemed inoperable and hence was treated with radiation therapy 9 years ago.

Case Report

A 40-year-old male presented with right-sided hip pain, numbness, and paresthesia along the right foot. Magnetic resonance imaging (MRI) of pelvis revealed a large expansile destructive cortical mass with associated soft-tissue component involving right sacrum, sacroiliac joint with extension along the right neural foramina. Based on these MRI findings, the possibility of a high-grade malignant tumor was considered, and hence the patient was sent for whole-body F-18 FDG PET/CT scan. Whole-body F-18 FDG PET/CT scan was acquired 45 min after intravenous injection of 300 MBq of FDG, to assess the extent of disease. The maximum intensity projection image (Figure 1 A-red arrow) revealed abnormal FDG uptake in sacrum and right sacroiliac joint. Fused transverse and coronal images of F-18 FDG PET/CT (B, C) revealed intense heterogeneous FDG uptake in right sacroiliac joint with SUVmax 28.05 and SUVmean 15.85. Corresponding transaxial and coronal CT images in bone window (D, E-white arrows) revealed cortical destruction and expansion of sacrum and right sacroiliac joint with associated large heterogeneously enhancing expansile soft-tissue mass infiltrating into the neural foramina of sacrum (F, G). Considering the intensity and pattern of FDG uptake in the destructive cortical lesion with soft-tissue mass, differential

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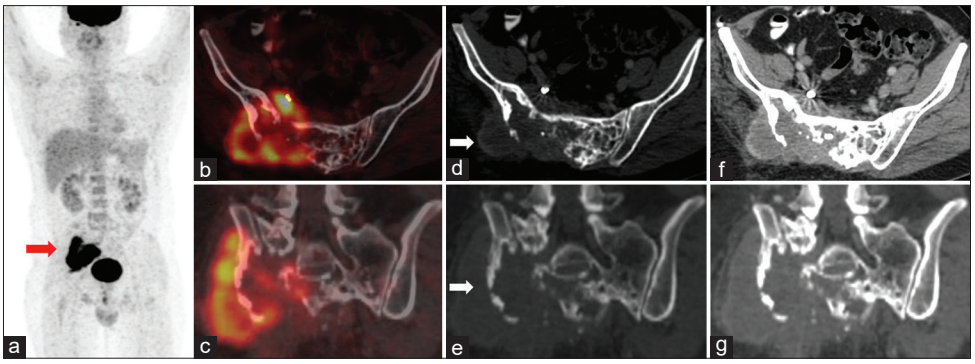

Figure 1: Maximum intensity projection (a- red arrow) shows intense uptake in right sacroiliac joint region. Fused positron emission tomography/computed tomography images (b and c) show intense heterogeneous FDG uptake in right sacroiliac joint which corresponds to destructive soft-tissue mass extending into neural foramina with cortical destruction and expansion seen on transaxial and coronal computed tomography images (d-g)

diagnosis of osteogenic sarcoma or soft-tissue sarcoma or primitive neuroectodermal tumors was made. On detailed interrogation, the patient gave a past history of recurrent GCT of sacrum. As the local tumor was deemed inoperable, he then was referred for radiotherapy and received a total dose of 54 Grays to the pelvis. Hence, collectively, history of local site irradiation and disease-free interval of 9 years and no prior history of sarcoma, these scan findings raised a high index of suspicion of radiation-induced sarcoma, which was confirmed on histopathology.

Discussion

Induction of secondary neoplasms is the most common delayed complication of external beam radiotherapy. Age at radiation exposure, dose, and type of radiation have a significant impact on the development of these secondary malignant neoplasms.[4] Radiation-induced sarcomas are rare malignant tumors involving bone and soft tissues as a result of exposure to high-dose radiation.[5] Diagnosis of radiation-induced sarcoma is made based on the fact that they are fast-growing tumors occurring at the site of irradiation with long latency period of 3–10 years, often metastatic at the time of presentation. Their incidence increases with longer survival resulting from local radiotherapy.[6] Clinically, these tumors are more aggressive as compared to other conventional soft-tissue sarcomas and often associated with worse outcome.[7] Hence, accurate diagnosis and early management of these tumors are important to improve outcomes. However, the diagnosis of radiologically isolated syndrome (RIS) and differentiation from recurrence of primary disease on conventional anatomical imaging is often challenging.[8] Locally advanced soft-tissue mass, cortical expansion, and adjacent bone destruction in the irradiated field of primary disease are the few nonspecific imaging findings which are seen on conventional imaging, whereas GCTs often show pure lytic lesion with thin nonsclerotic margins, trabeculations, and are not associated with bone destruction and soft-tissue components.[9,10] MRI is considered more specific for the diagnosis of RIS, as the normal marrow is replaced by fat after radiation therapy which appears hyperintense on T1-weighted images.[9,11] Repeated histological biopsies, though considered as the gold standard, often pose a diagnostic challenge due to sampling errors at difficult biopsy sites.[12] In such scenarios, noninvasive assessment of the functional status of the tumor can help inaccurate diagnosis. GCTs often show variable FDG uptake with SUVmax ranging from 2.5 to 10 due to the presence of modified macrophages, i.e., osteoclasts such as cells and mononuclear cells which can pose a diagnostic challenge.[13] However, intensity and pattern of FDG uptake directly correlates with tumor aggressiveness.[14] There is often low-grade patchy FDG uptake at the irradiated site due to chronic inflammation, however, intense uptake raises the possibility of a malignant etiology, and also provides a target for biopsy.[13,15] In our case, SUVmax of 28.05 and SUVmean of 15.85 which was significantly higher than the expected range for recurrent GCTs, helped in the differential diagnosis of radiation-induced sarcoma from recurrent GCTs. In addition, CT features of cortical expansion and bone destruction further confirm it to be a sarcoma at irradiated site. In our case of recurrent GCT, the patient had received high-dose radiation of 54 gray at a younger age. Along with these factors, high FDG uptake in the destructive cortical lesion with associated soft-tissue mass at the postoperative and postirradiated site involving right sacroiliac region seen on FDG PET/CT raised a high suspicion of radiation-induced sarcoma, which was later confirmed by histopathology. Thus, in patients with lesions in postradiation site, molecular imaging with PET/CT can confirm the diagnosis of RIS, when conventional imaging is equivocal.

Conclusion

F-18 FDG PET/CT has superior diagnostic accuracy in the evaluation of radiation-induced sarcoma.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and

other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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