

Case Report

Eosinophilic granuloma of the sacrum treated with radiation therapy: a case report

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Abstract

BACKGROUND CONTEXT: Eosinophilic granulomas (EGs) of the sacrum have been reported in fewer than 10 patients. Treatment algorithms for these tumors remain poorly defined; there are no reports of treating solitary sacral EG with radiation therapy (RT).

PURPOSE: This study aimed to describe the presentation, treatment, and outcome of sacral EG in an adult patient with intractable pain and radiculopathy, treated in a novel fashion with RT.

STUDY DESIGN/SETTING: The study design was a case report from a tertiary cancer referral center.

METHODS: Patient records, imaging, and pathology were reviewed.

RESULTS: A 35-year-old man received 20 Gy of radiation to his S1 EG lesion. He subsequently developed vertebra plana of S1 causing symptomatic L5–S1 stenosis, but 15 months after RT treatment was free of pain or tumor recurrence.

CONCLUSION: Radiation therapy is an effective treatment option for sacral EG causing severe axial pain and neural impingement. © 2015 Elsevier Inc. All rights reserved.

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Introduction

Eosinophilic granulomas (EGs) are benign bone lesions caused by Langerhans cell histiocytosis (LCH). Although EG in the mobile spine are relatively common (10%–15% of EG cases), reports of sacral EG are extremely rare [1,2]. We present a case of EG in an adult patient presenting with intractable pain and radiculopathy. The patient was successfully treated with a single course of radiation therapy (RT), a treatment not previously reported for solitary sacral EG. The patient subsequently developed vertebra plana of S1 causing L5–S1 foraminal stenosis.

Case report

A 35-year-old man with 2 months of progressive low back pain presented to the emergency department. His pain was initially 3/10 in severity but worsened to 10/10, requiring use of a walker; the pain radiated to both legs, and he experienced transient numbness of his left foot. He denied bowel



Fig. 1. Lateral radiograph of the lumbar spine, read as normal.

or bladder incontinence or lower extremity weakness, but did admit to a 7-kg unintentional weight loss and subjective fevers. His past medical history was unremarkable, and he had no family history of malignancies. He was initially evaluated by his general practitioner as well as a chiropractor, both of whom diagnosed sacroiliitis, and was prescribed non-steroidal anti-inflammatory drugs, which did not relieve his symptoms.

On initial examination at our institution, the patient had intact motor and sensory neurologic function, but had significant pain with ambulation that essentially confined him to bed. Initial radiographs were normal (Fig. 1). Magnetic resonance imaging (MRI) of the lumbosacral spine was performed, revealing a mixed solid and cystic lesion involving S1, with the solid components demonstrating T1 hypointensity, T2 hyperintensity, and contrast enhancement

(Fig. 2). The differential diagnosis based on imaging included aneurysmal bone cyst, hemangioma, and chordoma. Laboratory studies included an erythrocyte sedimentation rate of 16.0 mm/h (normal range 0.0–12.0 mm/h) and a C-reactive protein level of 25.3 mg/L (normal range 0–10 mg/L). Complete blood count and metabolic panels were normal. Dexamethasone and intravenous narcotics were administered for pain control, which were then transitioned to oral analgesics.

A computed tomography-guided bone biopsy was performed, with the CT images demonstrating an aggressive-appearing lytic lesion involving the majority of S1 (Fig. 3). Histology review showed epithelioid-like histiocytes with eosinophils and acute inflammatory cells. Immunohistochemistry demonstrated reactivity for vimentin, CD68, and CD1A; the latter finding in particular was highly consistent with eosinophilic granuloma (Fig. 4) [1,3]. A subsequent skeletal survey as well as a positron emission tomography scan demonstrated no other osseous or visceral lesions.

Spine surgical oncology, medical oncology, and radiation oncology were consulted. Treatment options were discussed with the patient, including surgical stabilization with tumor debriement or radiation therapy (RT). The patient elected to undergo external beam radiation, with a total of 20 Gy in 10 fractions of 2 Gy/d, delivered Monday to Friday over 14 elapsed days. At the conclusion of RT, the patient's pain had improved to 8/10, and he was able to walk.

Three months later, the improvement of his pain had plateaued to 4/10, and he also developed radiating pain from his right hip to his right foot. Because of his persistent pain, he was unable to return to work as a machinist. An MRI of the lumbosacral spine was obtained to further investigate the source of his persistent pain, which showed vertebra plana from an S1 compression fracture with mild bilateral L5–S1 neuroforaminal narrowing. He subsequently underwent an L5–S1 epidural corticosteroid injection, which significantly alleviated his pain. At 6 months post-RT, he was free of any

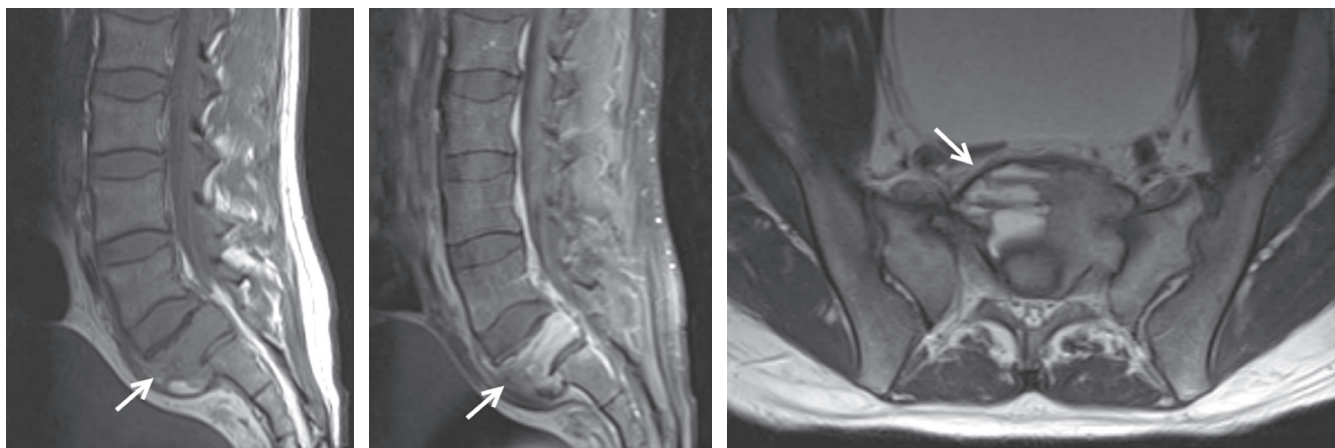


Fig. 2. Sagittal T1-weighted (Left) and post-contrast fat-saturated T1-weighted (Middle) MR images of the lumbar spine show an enhancing marrow replacing lesion (arrows) involving the majority of S1. An axial T2-weighted (Right) MR image demonstrates the mixed solid and cystic nature of the lesion with fluid-fluid levels (arrow).

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