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## Musculoskeletal

# Langerhans cell histiocytosis of bone in an adult: A case report

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### ABSTRACT

Langerhans cell histiocytosis (LCH) may clinically manifest in a variety of ways due to its ability to involve nearly every organ system. LCH may present as a single bone lesion, skin rash, or as invasive disseminated disease and occurs typically in the pediatric and adolescent population, affecting both males and females. Independent of its clinical presentation and severity, LCH lesions share the common histology of CD1a<sup>+</sup>/CD207<sup>+</sup> dendritic cells along with an inflammatory infiltrate, and, based upon improved scientific understanding, is now classified as a myeloproliferative neoplasm. We present a case report of an adult diagnosed with LCH of the pelvis.

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## Case presentation

A 54-year-old white man presented to an outside hospital emergency department for acute low back pain, flank pain, constipation, and left buttock pain radiating into his left thigh. He had no history of trauma to the area or an inciting event. His medical history was significant for psoriatic arthritis treated with Humira and a history of melanoma in situ. He underwent a computed tomography (CT) scan of the abdomen and pelvis with and without intravenous contrast along with a noncontrast CT of the lumbar spine, which revealed a 2.5-cm well-circumscribed osteolytic bone lesion involving the left ilium

with a narrow zone of transition. There was no discernible periosteal reaction or extraosseous soft tissue mass identified (Fig. 1). Laboratory workup (complete blood count, complete metabolic panel, and urinalysis) were within normal limits.

A bone scan was also performed and revealed abnormal focal intense radiotracer activity in the left iliac bone adjacent to the sacroiliac joint. Focus of radiotracer uptake in the right tibia was related to a prior fracture. Mild uptake in bilateral knees and ankles were also identified and may have been degenerative in nature or related to prior trauma (Fig. 2). The patient was subsequently referred to our institution for further evaluation.

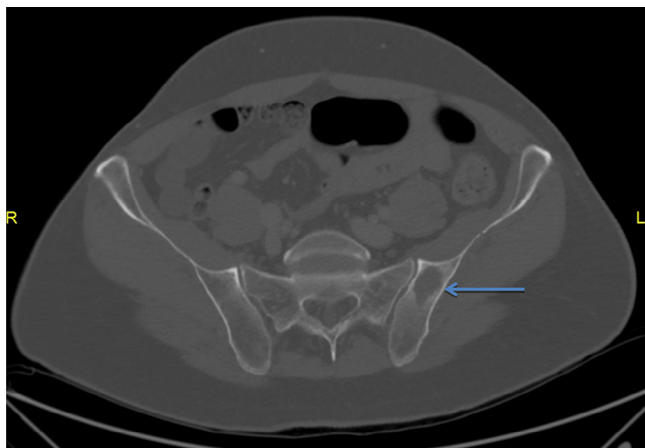
On presentation to our institution, physical examination was significant only for mild tenderness to palpation of his left ilium.

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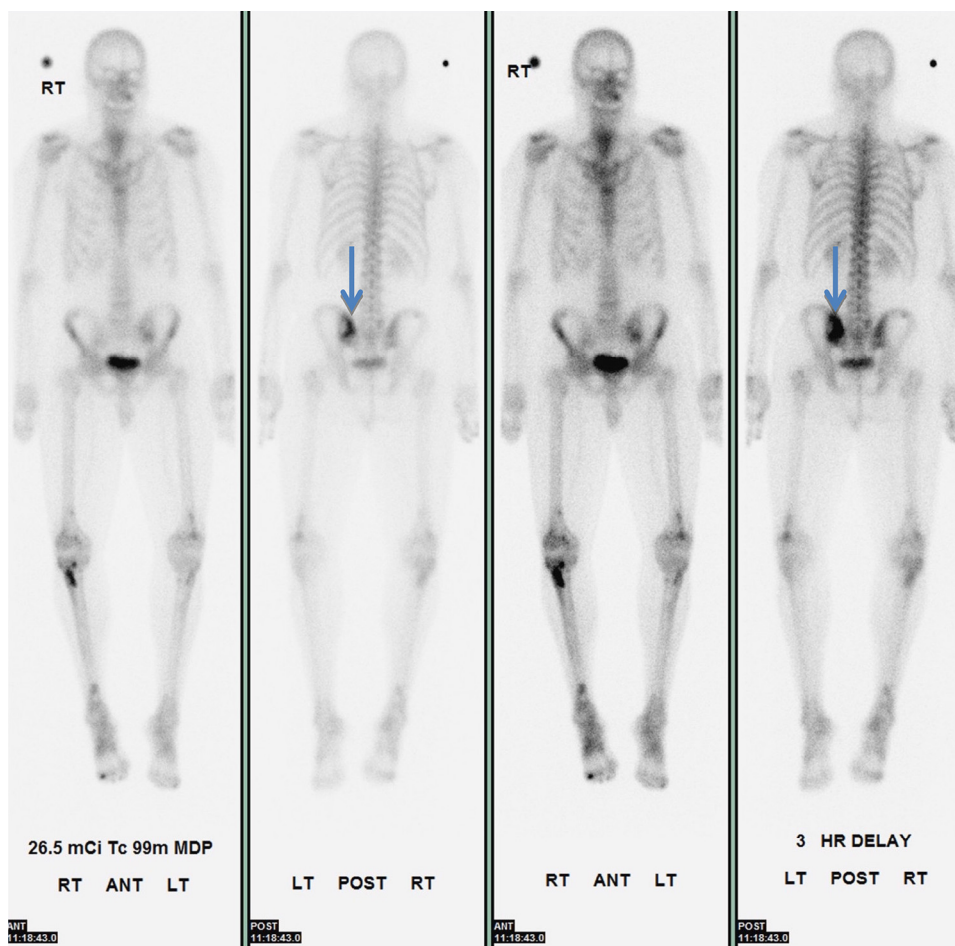


**Fig. 1 – Axial CT scan demonstrates a geographic osteolytic lesion of the posterior left iliac bone, with subtle areas of marginal sclerosis. Also demonstrated is intramedullary sclerosis of the adjacent bone, likely reactive in nature. CT, computed tomography.**

There was no palpable soft tissue mass, lymphadenopathy, or other site of pain.

The patient underwent a magnetic resonance imaging (MRI) of the pelvis which demonstrated an oval-shaped approximately 2.5-cm intramedullary lesion within the posterior left iliac bone centered at the level of the sacroiliac joint, corresponding to the osteolytic seen on prior CT scan. The lesion demonstrated slight T1 hyperintense signal to muscle, moderate T2 hyperintense signal and avid enhancement after the administration of intravenous contrast (most notable along the periphery). Edema-like signal on T2-weighted imaging adjacent to the lesion was suspected to represent reactive marrow edema (Figs. 3, 4, and 5).

Given the patient's age, the highest concern was for metastatic disease. Other considerations included plasmacytoma or osteomyelitis. The MRI findings were nonspecific and therefore a CT-guided biopsy with an 11-gauge Osteo-Site M2 bone biopsy needle was performed (Fig. 6). Two fine-needle and one core biopsy specimen were obtained. This revealed only minute fragments of bone and significant hemorrhage, thus an open biopsy was scheduled.



**Fig. 2 – Bone scan demonstrates increased radiotracer activity of the left iliac bone adjacent to the sacroiliac joint, corresponding to abnormality on CT. Intense radiotracer activity is also noted in the right proximal tibia from prior fracture and elsewhere within both knees and ankles, not relevant to the current discussion but possibly degenerative/posttraumatic in etiology. CT, computed tomography.**

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