Case Report

Infected primary non-Hodgkin lymphoma of spine

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Abstract

Primary bone lymphoma (PBL) comprises less than 5% of all malignant bone tumors and almost 7% of all extranodal lymphomas. Only 1.7% of all PBLs have been reported to involve the vertebrae. In our case, osteomyelitis was accidentally found during surgery, which might have resulted in the rapid collapse of vertebral body. This is the first report on primary lymphoma of the vertebrae with superimposed osteomyelitis in the English literature to the best of our knowledge. The patient reported here received anterior vertebrectomy and posterior interbody fusion with instrumentation for spinal instability. Tumor mass and the necrotic debris were removed. After the procedure, the patient received treatment with antibiotics and six cycles of chemotherapy. This case reminds us the possibility of hematologic seeding of bacteria in the tissue, especially with tumor necrosis. We suggest percutaneous needle aspiration for pathology and culture before making a decision whether or not to proceed with surgical decompression for fear of missing the occult bacterial infection.

Key words: Infected tumor, lymphoma, neurogenic claudication

Introduction

Primary lymphoma of the bone (PBL) is a rare neoplasm accounting for less than 5% of all malignant bone tumors and almost 7% of all extranodal lymphomas.[1] Occasionally, it may involve the vertebrae and encroach upon the nerve root, causing claudication and radicular syndrome mimicking spinal stenosis.[1] We report a case of PBL of the lumbar vertebrae and pelvis, initially presenting as back pain, lumbar radicular pain and numbness. During operation, pus-like fluid was noted and a positive bacterial culture was grown from the pus discharge and necrotic debris. To the best of our knowledge, this is the first reported case of PBL with superimposed bacterial osteomyelitis.

Case Report

A 71-year-old retired obstetrician was referred to our hospital due to severe pain over the lumbar region radiating to the right leg with numbness for 4 months. The patient reported that the pain had started in the proximal thigh down, moving to the lower leg after walking over 100 m, and was relieved by bending or sitting. He was treated with nonsteroidal anti-inflammatory medication, but without relief. On examination, tenderness and spasm on the lower back was noted. Mild weakness of the right ankle dorsiflexion was noted. Pain and numbness of the right medial leg region and a positive straight leg raise test on the right side were found. Except for an increased level of C-reactive protein (CRP; 9.25 mg/dL), other laboratory tests showed normal results for complete blood count, liver function, renal function, carcinoembryonic antigen, carbohydrate antigen 19-9, and electrophoresis of immunoglobulin.

A plain X-ray of the lumbar spine and sacrum showed a compression fracture of the L4 vertebral body and mixed osteoblastic–osteolytic changes to L4 [Figure 1a]. Osteoblastic changes were noted in the sacrum, ilium, and pubic bone [Figure 1b]. A bone scan with ^{99}Tc showed increased uptake by L4, L5, and the left-side pelvis. Magnetic resonance imaging (MRI) showed a compression fracture of L4 with an enhanced mass protruding into the spinal canal and neuroforamen [Figure 2], causing compression of the nerve root and cauda equina [Figure 3]. Positron emission tomography (PET) was used to further investigate the extent of the possible malignancy. Increased uptake of ^{18}F-labeled deoxyglucose (FDG) was noted over the lower lumbar region and left-side bony pelvis, consistent with the MRI findings. There was no evidence of the dissemination of the malignancy from other parts of the body.

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Anterior decompression of L3–L5 and anterior fibular strut interbody fusion with right anterolateral retroperitoneal approach [Figure 4] were performed to relieve the patient's aggravated back pain and neurological symptoms. Pus-like fluid was noted when the pseudocapsule of the tumor was opened. Specimens were taken for Gram's stain and culture. Also, tumor infiltration of L4 and L5 was observed intraoperatively. A grayish tumorous mass was found to occupy the L4 vertebra, protruding into the paraspinal region and compressing the dural sac and neuroforamen. During the surgical decompression and curettage of the tumor, necrotic tissue with pus discharge was retrieved from the L4 vertebra. Because of the extent of the destruction of the L4 vertebra, a corpectomy of L4 was performed. One week later, a second-stage posterior instrumentation with pedicle screw system from L2 to S1 and posterolateral fusion were done [Figure 4]. Allogenous fibula strut graft and bone substitute were used for interbody fusion. A rotational table was used to assist positioning the patient in a prone position to avoid displacement of the fibula allograft.

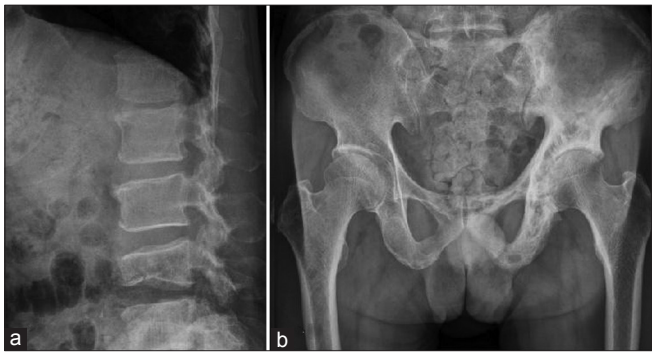

Figure 1: (a) Compression fracture of the L4 vertebral body and mixed osteoblastic–osteolytic changes to L4. Loss of lumbar lordotic curve and decreased L4/5 disc height were noted. (b) Osteoblastic changes were noted in the sacrum, ileum, and pubic bone

Figure 3: Magnetic resonance scans T1WI (a) and T2WI (b) of the lumbar spine, showing lytic lesion of L4 vertebrae, bone marrow edema, with erosion through endplates and narrowing of L4/5 disc space. There is an extradural obstruction at the L4/5 interspace and a paraspinal soft tissue mass

Pathological and immunological analyses showed large and monotonous tumor cells infiltrating the stromal tissue, which were positive for marker CD20 and negative for CD3 [Figure 5]. Three sets of bacterial culture of the tissue fluid and tumor debris resulted in the growth of methicillin-susceptible Staphylococcus aureus. Also, acid-fast stain and tuberculosis polymerase chain reaction (PCR) were done to rule out tuberculosis of bone. The result came out as negative. These studies confirmed the diagnosis of diffuse large B-cell lymphoma with superimposed osteomyelitis.

The patient had immediate relief of his radicular pain after the operation. He was treated with 2 g oxacillin intravenous injection every 4 hours for 6 weeks and switched to oral ceflexin 500 mg every 6 hours for 2 weeks. After the treatment with antibiotics, the CRP level

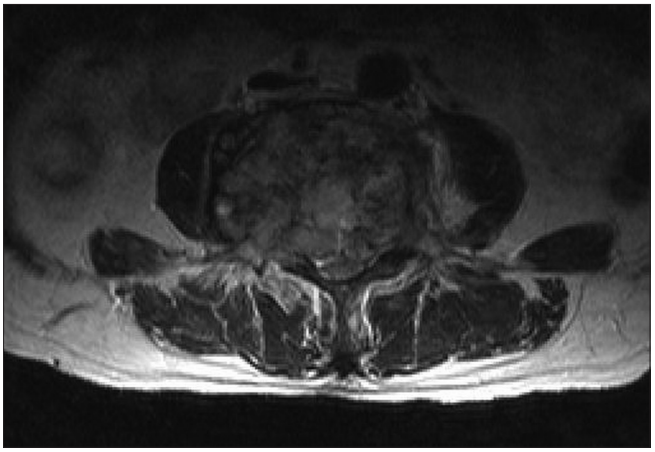

Figure 2: Enhanced mass with heterogeneous signal intensity originating from L4 vertebral body protruded into the spinal canal and neuroforamen, causing compression of the nerve root and cauda equina

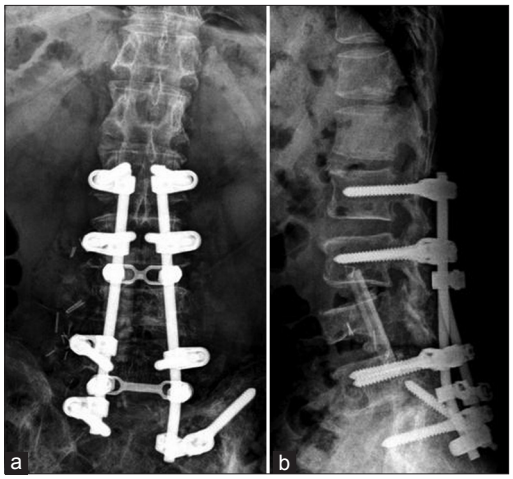

Figure 4: Anteroposterior and lateral radiographs of lumbar spine 7 months after surgical intervention: (a) anterior decompression with vertebrectomy of the fourth lumbar vertebral body and interbody fusion with structural fibula allograft and autologous bone graft; (b) posterior stabilization and fusion of L2–S1; note reduction of deformity and restoration of lordotic curve

came down to 5 mg/dL on the 7th postoperative day. The value gradually came down to 2 mg/dL on the 14th postoperative day. The value stayed at a stable value of 1.5 mg/dL in the 3rd and 4th postoperative weeks. With a stable surgical wound and stable CRP value, the treatment was followed by six cycles of chemotherapy including rituximab, cyclophosphamide, epirubicin, vincristine, and prednisolone (R-CEOP). The patient resumed nearly normal life 4 months after surgery, with only slight weakness of right ankle dorsiflexion.

A post-therapy assessment was made with PET/CT, 6 months after the operation. The previously noted FDG-avid region, including the L4 vertebra, sacrum, and left-side pelvis, was not detectable in the post-therapy study. The patient was still in remission 20 months after presentation. No systemic involvement has been noted as of the date of publication.

Discussion

PBL is defined as a lymphoma of the bone without any systemic involvement within 6 months of the first diagnosis. The incidence is less than 1% of that of primary non-Hodgkin lymphoma.[1-3] The lesion is commonly located in a short bone, such as the ilium, scapula, or vertebra.[3] PBLs involving a single vertebra comprise 1.7% of all PBLs.[4]

Spinal osteomyelitis is estimated to occur in approximately 1 per 100,000 persons annually.[5] The predominant organism in almost all studies is S. aureus, accounting for approximately 40–80% of all spinal infections.[6] A distant focus of infection provides an infective nidus from which bacteria spread via the bloodstream to the spinal column. The skin and genitourinary tract are common antecedent sites.[7] Our patient presented with osteomyelitis, with no history of trauma or previous surgery. The hematological spread of microorganisms to areas of tumor necrosis may be the basis of the infection.

Both PBL and spinal osteomyelitis can present as gradually progressive back pain that increases with movement . Fever is present in approximately 50% of patients with spinal osteomyelitis. A differential diagnosis may be difficult at initial presentation.

The diagnosis of PBL relies on both radiological and pathological studies, and it can be easily missed on plain radiographs.[8,9] In our patient, the lesions appeared osteoblastic in the pelvic and iliac bones, which can lead to a misdiagnosis of metastatic disease in the first impression. In the L4 vertebral body, the lesion appeared to be a mixed osteolytic–osteoblastic lesion. On MRI, the lesion of our patient appeared like most other lymphoid malignancies, which produce isointense signals on T2-weighted images.[8]

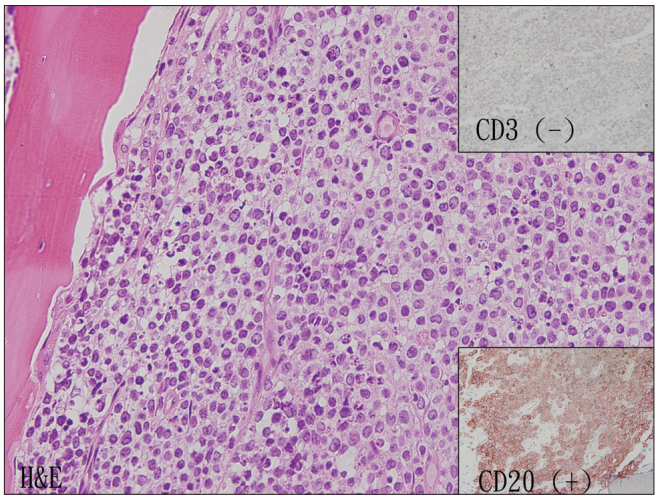

Figure 5: The section shows a picture of malignant lymphoma, B cell linage, characterized by dyscohesive, large, and monotonous tumor cells infiltrating within the stromal tissue with irregular nuclear membrane and focal bony erosion. The insets show that these tumor cells are immunoreactive to CD20 and negative for CD3 (magnification ×400)

Because most metastatic cancers or sarcomas give higher signals on T2-weighted images, this may be a way to distinguish lymphoid cancer from the other pathologies.[8]

The treatment for PBL, which usually consists of chemotherapy, radiotherapy, or both, is potentially curative.[9] Although PBL seemed to be the major medical problem of this patient, osteomyelitis, if not treated in time, can cause spinal cord infection and epidural abscess, potentially resulting in paralysis within days, especially if the patient is treated immediately with chemotherapy and radiotherapy.[9]

Surgical decompression and stabilization can be performed to treat the mechanical compression symptoms and spinal instability due to the vertebral body destruction attributable to both PBL and osteomyelitis. There have been debates regarding the use of allograft or autograft for spinal fusion. Daniel *et al.* reported an undetectable difference in the rates of recurrent infection between autografts (5.9%) and allografts (5.3%).[10]

A recent literature review of the treatment of PBL recommended combined radiotherapy and chemotherapy, which produced a 95% 5-year overall survival rate compared with 70% in patients treated with radiotherapy alone and 81% in patients treated with chemotherapy alone.[11] Both chemotherapy and radiotherapy were important in the treatment of lymphoma. Primary localized lymphoma is generally treated with debulking surgery accompanied by local radiotherapy and chemotherapy. However, in the context of superadded localized infection, combined radiotherapy and chemotherapy may further

compromise the patient's immune system and delay wound healing. Considering the patient's age, proximity of two major surgeries, and multiple instrumentations, treatment team decided to postpone the chemotherapy until good control of infection and wound healing was achieved.

This case is unique in the literature because it is the first reported case of PBL with superimposed osteomyelitis, with no history of trauma, previous surgery, or biopsy. We recommend extra caution in the treatment of patients with PBL of the vertebrae to avoid overlooking potential osteomyelitis in immunocompromised patients. We may consider percutaneous needle biopsy and culture to avoid missing occult infection.

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