

Case Report

Bone Involvement in Hodgkin's Disease at Presentation: A Series of Three Case Reports with a Brief Review

Farhat Aziz Khan, Shad Salim Akhtar, Muhammad Kamil Sheikh

Department of Medical Oncology, King Fahd Specialist Hospital, Buraidah, Al-Qassim, Saudi Arabia

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ABSTRACT

Most patients with Hodgkin's lymphoma present with progressive painless lymphadenopathy. Very few patients have presented with bony involvement at the time of diagnosis. We report on three cases of Hodgkin's lymphoma who presented with signs and symptoms of bone involvement in addition to other clinical features of

the disease. On staging workup, all the three patients were found to have bony lesions at the time of diagnosis, confirmed by imaging studies. All patients were treated with chemotherapy and radiotherapy directed at their bony lesions.

KEYWORDS: bony lesions, Hodgkin's lymphoma, painless lymphadenopathy

INTRODUCTION

The most common presentation in Hodgkin's disease is progressive painless lymphadenopathy. Although bone involvement is frequent during the course of disease, an onset with bone involvement and its symptoms is extremely rare. We present here three cases of Hodgkin's disease, one male and two female, diagnosed at King Fahd Specialist Hospital, Al-Qassim, Saudi Arabia, over a period of three years. All the three patients were young and presented with signs and symptoms of bone involvement in addition to other clinical features of Hodgkin's lymphoma. Two patients presented with backache and one with pain in the left hand and left arm. Diagnosis was confirmed histopathologically in all patients. On staging workup, all the three patients were found to have bony involvement at the time of diagnosis confirmed by imaging studies. Diagnosed as stage IVB disease, all of them received treatment in the form of chemotherapy and radiotherapy directed at their bony lesions.

Case - 1

A 19-year-old female presented with a four week history of backache, irregular fever and weakness. During this period, she also noticed small multiple swellings on both sides of her neck and lost some weight as well. Her back pain was moderately intense without any radiation or motor power weakness. There was no history of any urinary, respiratory or gastrointestinal manifestations. Also, she did not give history of any definitive treatment. On examination, she was ill-looking,

pale and febrile. Multiple bilateral cervical and right axillary lymph nodes were palpable. Her back examination revealed a spinal gibbous and tenderness over lower dorsal and lumbar spines. Abdominal examination showed mild hepatospleno-megaly and rest of the systemic examination was normal.

Laboratory investigations revealed an Hb of 7 gm/dl and a high LDH level. Rest of the parameters was normal. Her chest X-ray (CXR) showed bilateral hilar and paratracheal lymphadenopathy and an X-ray of the dorsolumbar spine showed a compression fracture of L1 vertebra (Fig. 1). A CT scan of the neck, chest and abdomen showed bilateral pretracheal, paratracheal, subcarinal and right hilar nodes. Multiple para-aortic and inter-aortocaval lymph nodes were also seen. The liver and spleen were normal. A MRI of the spine confirmed a wedge shaped compression fracture of L1 vertebra with bone destruction. Post contrast MRI showed an adjacent para-spinal enhanced soft tissue invasion and prevertebral lymph node mass at the level of L1 without epidural invasion or cord involvement; there was also post contrast enhancement in the bodies of T9, T12, L2 and L5 vertebrae (Fig. 2). Radioactive isotope bone scan suggested increased uptake in L1, L4 and L5 and bilateral greater trochanters. Bone marrow aspiration and biopsy did not confirm any marrow infiltration. Fine needle aspiration of cervical lymph node suggested Hodgkin's disease which was subsequently confirmed by biopsy. Diagnosed as stage IVB Hodgkin's disease, she was treated with chemotherapy (ABVD x 6) and simultaneous

Address correspondence to:

Dr. Farhat Aziz Khan, Advanced Medical And Dental Institute, Universiti Sains Malaysia Suite 141, Eureka Complex, 11800 USM, Penang, Malaysia. Tel:04-6532738/017-4691636, Fax:04-6532734, E-mail:fkhanmurad@hotmail.com



Fig. 1: X-Ray of Lumbar spine (lateral view) showing compression fracture of L1 vertebrae

radiotherapy to her spine. The patient was discharged after her first cycle of chemotherapy and advised to follow up for further treatment.

Case - 2

A 28-year-old Saudi male patient was admitted with history of backache, shortness of breath, generalized weakness and loss of weight for one month. He also noticed bilateral multiple swellings in his neck and there was associated history of night sweats and occasional low-grade fever. On examination, he was dyspneic at rest and had bilateral lymphadenopathy in supraclavicular, jugulodigastric, subclavicular and axillary regions. The nodes were rubbery and the biggest one over left jugulodigastric region was 5 cm x 2 cm in diameter. Chest examination revealed diminished breath sounds over left hemithorax with stony dull note on percussion suggestive of pleural effusion. Cardiovascular and nervous system examination was normal. Abdominal examination showed hepatosplenomegaly. Musculoskeletal examination revealed tenderness over the lumbar area.

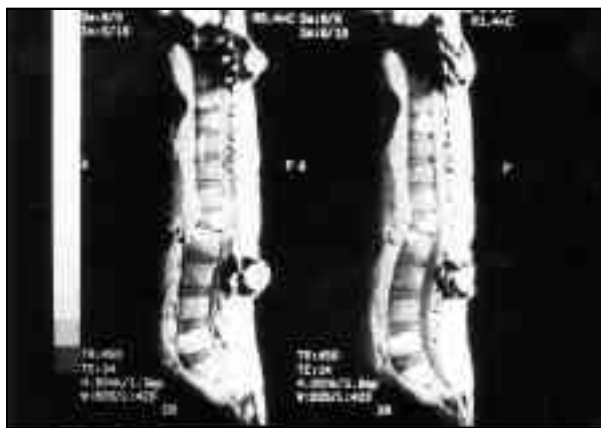


Fig. 2: Spinal MRI showing a wedge shaped compression fracture of L1, post contrast enhancement in the bodies of T9, T12, L1, L2 and L5 vertebrae. There is no epidural invasion or cord compression. Also shown, is the spinal gibbous.



Fig.3: CT scan of abdomen showing retroperitoneal, paraaortic lymphadenopathy with infiltration of right psoas area and right border of second lumbar vertebrae

Investigations revealed a WBC count of $28,000 \times 10^9/L$ with 91% neutrophils and 3.3% lymphocytes. Hemoglobin was 11.2 gm/dl and the platelet count was normal. Serum chemistry was unremarkable except for low total proteins (40.6 gm/l). Lymph node biopsy revealed the diagnosis of a grade-II nodular sclerosis type of classical Hodgkin's lymphoma. Bone marrow aspiration did not reveal any bone marrow infiltration. His CT scan brain, neck, chest, abdomen and pelvis showed bilateral cervical lymphadenopathy, massive left-sided pleural effusion with collapse of left lung and evidence of retroperitoneal and para-aortic lymphadenopathy with hypodense necrotic mass infiltrating in the region of the right psoas muscle, 2nd and 3rd lumbar vertebrae and medial aspect of the right kidney (Fig. 3 and 4). Diagnosed as Stage-IVB Hodgkin's disease, he was started on chemotherapy (ABVD) and simultaneous radiation therapy to his spine. After receiving his first cycle of chemotherapy the patient was asymptomatic and was discharged with advice to follow up for further chemotherapy.



Fig. 4: Reconstructed CT scan image showing destroyed lumbar vertebral body



Fig. 6: Periosteal apposition of postero-lateral and medial aspect of middle 3rd of left femur

Case - 3

A 16-year-old young girl presented with a complaint of pain, mainly in left hand and left arm and a fever of two week duration. There was no history of any visible swelling, weight loss or sweating. On physical examination, she was looking well with stable vital signs. She had left supraclavicular lymphadenopathy. A musculoskeletal



Fig. 5: X-ray left hand showing spongiosclerosis with periosteal apposition of 2nd and 3rd metacarpal bones

system examination revealed an inflammatory swelling over the dorsum of her left hand and tenderness of the left arm below elbow joint and left mid thigh. Other systemic examination was unremarkable.

CBC and chemistry was normal. A CXR revealed haziness over left apex (left paratracheal lymphadenopathy). Abdominal USG was normal and CT scan chest, neck and abdomen showed left cervical and paratracheal lymphadenopathy. Limb X-rays revealed multiple sclerotic bony lesions - viz., (i) spongiosclerosis with periosteal apposition affecting 2nd and 3rd metacarpal bones of the left hand (Fig. 5), (ii) periosteal apposition of middle third of right radius with focal spongiosclerosis, and (iii) periosteal apposition of the postero-lateral and medial aspects of the middle 3rd of left femur (Fig. 6). Excisional biopsy of the left cervical lymph node was done and histopathology was suggestive of Hodgkin's disease, nodular sclerosis. Diagnosed as stage IVB with bone involvement, this patient was started treatment on chemotherapy (ABVD x 6 cycles) and after completing full treatment the patient was in complete remission.

DISCUSSION

Hodgkin's disease usually presents with nodal and visceral enlargement. Bone involvement in Hodgkin's disease at the time of presentation is extremely rare^[1,2]. Less than 20 such cases have been reported in the literature. Kaplan *et al*^[3] reported only four out of 334 cases of Hodgkin's disease to have radiologically demonstrable skeletal involvement at the time of initial diagnosis. Gross *et al*^[4] reported two cases of primary Hodgkin's disease of the bone. Gall and Mallory^[5] did not find a single case with initial bone involvement out of 229 cases of Hodgkin's disease studied by them. Most of the cases with bone involvement at initial presentation have been adolescents and few adults, which is comparable with our case reports.

Osseous involvement in Hodgkin's disease occurs through hematogenous spread or direct spread from a contiguous involved lymph node. The presenting complaints of bone involvement are usually insidious onset of pain with associated weight loss and malaise. Radiologic evaluation reveals osteosclerosis alone, osteolysis alone or a combination of two. Osteolytic lesions are poorly defined and are associated with periostitis in approximately one third of cases^[6]. Technicium 99 bone scan may disclose other clinically unapparent sites of bone involvement. The common sites are spine, pelvis, ribs, femur and sternum^[7].

For isolated bone lesions radiotherapy is quite effective and long-term disease-free survival in such patients has been reported^[8]. Patients with pelvic or spinal bone involvement are unlikely to

survive for long because their response to cytotoxic therapies is relatively poor^[9]. In the present series, treatment consisted of chemotherapy (ABVD x 6 cycles) and radiation therapy directed at the bony lesions.

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