Bone Infarcts in a Case of Chronic Myeloid Leukemia: Chronic Phase

Sanjeev Kumar Sharma · Tulika Seth ·
Pravas Mishra · Manoranjan Mahapatra

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Dear Editor,

Infarcts are commonly seen in spleen, and rarely in myocardium and brain, in patients of chronic myeloid leukemia (CML). Bone infarcts, manifesting as severe debilitating pain as an initial presentation of CML have not been reported. We present a case of CML in chronic phase whose initial presentation was severe bone pains with inability to walk. Tc-99m MDP bone scan revealed heterogeneous areas of bone infarct involving long bones of both upper and lower limbs. She was treated with Imatinib and Ecosprin. She became complete pain-free in 4 months. This case highlights the unusual presentation of CML-CP and complete recovery with standard therapy.

A 32-years old woman presented with diffuse bony-pains involving bilateral upper and lower limbs for 3 weeks duration. She was not able to perform her daily routine activities because of severe pain (ECOG performance status 3). Clinical examination revealed mild pallor and splenomegaly (spleen size 8 cm below costal margin). Hemogram revealed hemoglobin 9.4 g/dl, total leukocyte count $136 \times 10^9/l$, platelet count $588 \times 10^9/l$ and differential count-neutrophils 16%, lymphocytes 22%, myelocytes 24%, metamyelocytes 34%, basophils 3% and blasts 1% (Sokal, Intermediate risk 0.83). Her biochemical profile revealed blood urea 36 mg/dl, creatinine 1.1 mg/dl, uric acid 4.2 mg/dl, calcium 8.6 mg/dl and bilirubin 0.8 mg/dl. Her leukocyte alkaline phosphatase (LAP) score was low

and RT-PCR for BCR-ABL was positive for p210. She was diagnosed as a case of chronic myeloid leukemia- Chronic phase (CML-CP). Radiographs of limb bones were normal. Tc-99m MDP bone scan revealed heterogeneous increased radiotracer uptake in limb bones (involving bilateral humeri, forearm bones, femurs), suggestive of multiple bone infarcts. Rest of the skeleton was normal. She was started on hydroxyurea 500 mg thrice daily, allopurinol 100 mg thrice daily, imatinib mesylate 400 mg once daily and ecosprin 75 mg once daily. She was given analgesics (paracetamol and tramadol) for bone pains. Hydroxyurea and allopurinol were stopped after 2 weeks and imatinib and ecosprin continued. Patient achieved complete hematological response after 2 months of imatinib therapy but continued to have limb pains for 4 months though with decreased intensity. Pain completely subsided 5 months later (ECOG performance status 0). Repeat bone scan performed at 5 months revealed completely normal bone scan. Quantitative PCR for BCR-ABL at 15 months revealed major molecular response.

CML accounts for approximately 15 to 20% of leukemias in adults. The clinical findings at diagnosis of CML-CP vary among reported series. Twenty to 50% of patients are asymptomatic, with the disease first being suspected from routine blood tests [1, 2]. Among symptomatic patients, systemic symptoms such as fatigue (34%), weight loss (20%), abdominal fullness (15%) and bleeding episodes due to platelet dysfunction (21%) are common [2]. Abdominal pain and discomfort may include left upper quadrant pain and early satiety due to the enlarged spleen with or without perisplenitis and/or splenic infarction. Tenderness over the lower sternum, due to an expanding bone marrow, is sometimes seen. Acute gouty arthritis may also present due to overproduction of uric acid [3]. Other frequent findings include splenomegaly

S. K. Sharma (✉) · T. Seth · P. Mishra · M. Mahapatra
Department of Hematology, IRCH, All India Institute of Medical
Sciences, 1st Floor, New Delhi, India
e-mail: sksanjeev13@yahoo.com

(present in 48–76%), anemia (45–62%), white blood cell count above $100 \times 10^9/l$ (52–72%) and platelet count above 600×10^9 (15–34%) [1, 2]. Involvement of extra-medullary tissues such as the lymph nodes, skin, and soft tissues is generally limited to patients with blast crisis. Although thrombocytosis is common, thrombosis is not seen [2]. Other rare manifestations include splenic rupture [4] and priapism [5]. The radiological changes studied in CML include diffuse osteoporosis, focal osteolytic and osteoblastic lesions, chloromas and arthritis [6]. Bone pains are one of the common presentations of hematological malignancies [7] and are usually due to marrow infiltration by leukemic cells causing expansion of bone marrow. Bone infarction with severe pain as initial presentation has never been reported in CML-CP, though a case of CML developing bone infarction and progressing to myelofibrosis has been documented [8]. Our patient presented with diffuse debilitating bone pains due to multiple bone infarcts which is an unusual presentation of CML-CP. The patient responded to the usual treatment with complete disappearance of bone lesions and complete clinical recovery.

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