Abstract: Primary bone lymphoma represents approximately 3% of all malignant bone tumours and 5% of all non-Hodgkin lymphoma. A 65 yrs old gentleman from Eastern India presented with 3 month history of continuous dull aching pain in left iliac region radiating to left thigh. Initial examination revealed a 3 cm tender swelling with indistinct margin localized over the lateral portion of the left iliac crest region. A needle biopsy was performed. The histopathological analysis showed non-specific chronic osteomyelitis. The culture sensitivity showed no growth. In view of biopsy report he was started on broad spectrum antibiotics for 8 weeks. Patient came back after 8 months for follow up. He was pale, cachexic and had significant loss of weight. On examination the swelling had increased to size of 10 cm, soft tissue mass present in left iliac region extending up to inguinal region. A repeat open biopsy done which revealed scanty, high grade B cell Non Hodgkin lymphoma, biopsy left ilium. Medical oncology opinion was obtained and the tumor staged as Ann Arbor Ia Extra nodal, with risk stratification of low-intermediate. To our knowledge, this is the first report of an innominate bone lymphoma in India. This has been reported due to the unusual location of this tumor, the ambiguity in clinical symptoms and the diagnostic dilemma thus caused.

Keyword: lymphoma of pelvis, osteomyelitis of pelvis

INTRODUCTION

Primary bone lymphoma represents approximately 3% of all malignant bone tumours and 5% of all non-Hodgkin lymphoma (1). Most occurrences present similar characteristics such as a slightly greater frequency in men usually in fifth and sixth decade. The common site is long bone (50%), followed by the axial skeleton (44%). The incidence of lymphoma of innominate bones are very rare (2). On imaging the pelvic lymphoma can mimic other tumorous lesion and infection. Hence it’s necessary to differentiate it from these lesion for good outcome. We present this case of primary pelvic bone lymphoma due to its rarity and diagnostic dilemma.

CASE REPORT

A 65 yrs old gentleman from Eastern India presented with 3 month history continuous dull aching pain in left iliac region radiating to left thigh. He did not have constitutional symptoms like fever, weight loss and loss of appetite. He had no known medical co-morbidities and family history was not contributory. Initial examination revealed a 3 cm tender swelling with indistinct margin localized over the lateral portion of the left iliac crest region. There was increase in temperature and erythema over the swelling with no involvement of skin or presence of dilated veins. Movements of left hip were restricted due to pain. General examination was unremarkable except for pallor. There was no localized or generalized lymphadenopathy. Blood investigations revealed the normal haemogram, mild elevated ESR and CRP levels. Radiograph revealed an osteolytic lesion with indistinct margins, moth eaten appearance and narrow transitional zone (Fig 1). Bone scan showed isolated uptake at the left ilium. In view of age and location differential diagnosis included primary bone tumor and indolent infection. A needle biopsy was performed.

The histopathological analysis showed non-specific chronic osteomyelitis. The culture sensitivity showed no growth. In view of biopsy report he was started on broad spectrum antibiotics for 8 weeks. Patient came back after 8 months for follow up. He was pale, cachexic and had significant loss of weight. On examination the swelling had increased to size of 10 cm, soft tissue mass present in left iliac region extending up to inguinal region. The left lower limb was edematous. There was no localized lymphadenopathy. Plain radiograph of pelvis showed a permeative lesion with significant periosteal reaction over the left iliac crest (Fig 2). The chest radiograph, computed tomography of chest was negative for any lesion. A repeat open biopsy done which revealed scanty high grade B cell Non Hodgkin lymphoma, biopsy left ilium. Medical oncology opinion was obtained and the tumor staged as Ann Arbor Ia – Extra nodal, with risk stratification of low-intermediate. He was planned on combined treatment modality with chemotherapy and radiotherapy.

DISCUSSION

Primary Lymphoma of the bone usually presents with constant pain, with no relief on rest. Often a palpable mass is present due...
to soft tissue extension of the tumor. Swelling, pathologic fracture, cord compression and systemic “B” symptoms (i.e., fever, weight loss, night sweats) may also be present at the time of diagnosis. Often, it may be several months before the patient seeks medical attention for these symptoms, causing a diagnostic delay.

Limb et al showed there is mean delay of eight month in the diagnosis of lymphoma of the bone (2). Our patient presented with symptoms of bone pain and palpable mass. However, the paucity of systemic ‘B’ symptoms and relative rarity of lymphoma in the ilium made us consider other differential diagnosis after clinical examination leading to a diagnostic delay of ten months from initial symptoms.

Primary lymphoma of bone are uncommon. In a series of 340 consecutive untreated patients, only four patients had skeletal involvement (1). Skeletal involvement of primary lymphoma is mainly limited to the long bones and axial skeleton, with femur being the most common site of occurrence. Though intra-pelvic, non-osseous lymphoma has been reported, involvement of the nominate bones has been described sparingly, with few case reports in world literature (3). The diagnosis of lymphoma presenting as a primary bone lesion can cause a diagnostic dilemma because neither the clinical features nor the radiological appearance are pathognomonic.

Radiologically the bone lesion are usually multiple. The lesion have no characteristic features and may be lytic (75%), blastic (13.6%) or mixed (5.8%) (4). CT scans; MRI and PET scans though helpful in assessment and staging of the tumor are neither pathognomonic nor diagnostic. The radiological picture in our patient at first presentation demonstrates a lytic, radiolucent lesion, cortical erosion which represents the possibility of infection and was correlated with the biopsy report. Whereas the radiological picture on second presentation after one year the lesion represents the permeative lesion with defined margin suggestive of the malignant tumor. Hence there is the dilemma in the diagnosis with radiological feature of the lymphoma. Closed or Open biopsy followed by histopathological examination is the gold standard diagnostic investigation. The majority of primary bone lymphomas are pathologically diagnosed as diffuse large B cell lymphoma (DLBCL), with histological features of infiltration of lymphoid cell with prominent nucleoli. Indolent lymphomas, such as follicular lymphoma, small lymphocytic lymphoma and marginal zone lymphoma may present as PLB. Highly aggressive subtypes, such as Burkitt or lymphoblastic lymphoma are rare. In view of the patient’s age, ambiguity in the clinic-histological findings, the patient was followed up clinically for ten months. With increasing symptoms, a repeat biopsy was performed which showed findings suggestive of diffuse large B cell lymphoma (DLBCL).

The lag time delay poses a diagnostic difficulty thus emphasizing the need for high degree of suspicion when clinical and histopathological findings do not tally. To our knowledge, this is the first report of an innominate bone lymphoma in India. This has been reported due to the unusual location of this tumor, the ambiguity in clinical symptoms and the diagnostic dilemma thus caused.

**FIG 1 :** Osteolytic lesion with indistinct margins, moth eaten appearance, cortical erosion and narrow transitional zone

**FIG 2 :** Pelvis X-ray showing a permeative lesion with significant periosteal reaction over the left iliac crest

**BIBLIOGRAPHY**
