

# Follicular Lymphoma with Musculoskeletal Presentation Mistaken for Osteomyelitis: Case Report



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## Abstract

Primary osseous/skeletal lymphoma is a rare neoplasm of lymphoid lineage which presents with single or multifocal lesions in a single bone without any definite nodal or extranodal involvement at the presentation or any evident distant metastasis for six months after initial diagnosis. They usually present with a single lesion with permeative pattern of bone destruction. As compared to it, multifocal primary bone lymphomata are less common. Diffuse large B cell type represents most of the Primary bone lymphoma. The follicular subtype variety is considered rare to present with musculoskeletal manifestation. Here we are discussing a case of follicular lymphoma in a young male who presented with pain and swelling in left arm and was treated initially for osteomyelitis and later on diagnosed to have lymphoma after a lag of eight months. We aim to highlight on keeping a high index of suspicion of malignancy in cases with equivocal response when treated for osteomyelitis.

**Keywords:** Primary bone lymphoma (PBL); Osteomyelitis; Follicular lymphoma (FL)

## Introduction

Lymphoma is a malignant neoplasm of lymphoid lineage arising from either B or T type lymphocytes or by natural killer (NK) cells. It is divided into hodgkins and non-hodgkins types. According to World Health Organization (WHO) classification of soft tissue and bone tumors [1,2] in 2020, primary bone lymphoma is characterized as neoplasms of malignant lymphoid cells presenting with one or more bone lesions without any nodal or extranodal involvement. Primary bone lymphoma is considered rare and accounts for about 7% of primary malignant bone tumors, approximately <1% of all lymphomas and about 3-7% of extranodal lymphomas [3-5]. Of all the subtypes, diffuse large B cell (DLBCL) and not otherwise specified (NOS) subtypes are common and follicular subtype of primary bone lymphoma is considered rare.

## Case Report

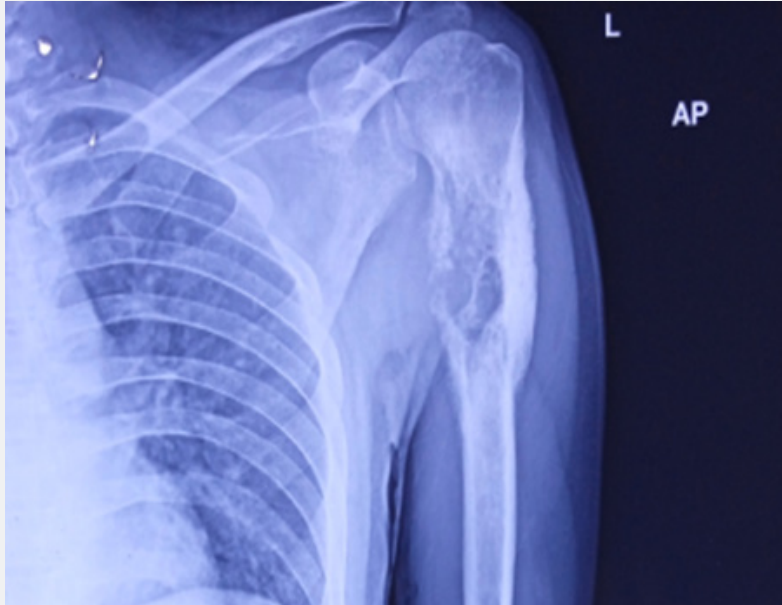
A 27-year-old young male reported to Orthopedics department of this hospital with complaint of painful swelling in left arm for eight months for which he underwent a surgery

in another hospital however without any significant relief in the post-operative period. On reviewing his documents, he was found to have been managed as a case of chronic osteomyelitis of humerus(left) treated with IV antibiotics, bone debridement and bone grafting performed seven months back. The initial HPE specimen was reported inconclusive. The index radiograph reported an ill-defined irregular marginated osteolytic medullary lesion in upper shaft with associated permeative bone destruction in subjacent medullary cavity with exuberant cortical thickening. During initial convalescence after surgical debridement, he had a pathological fracture for which an external fixation was also done six months back.

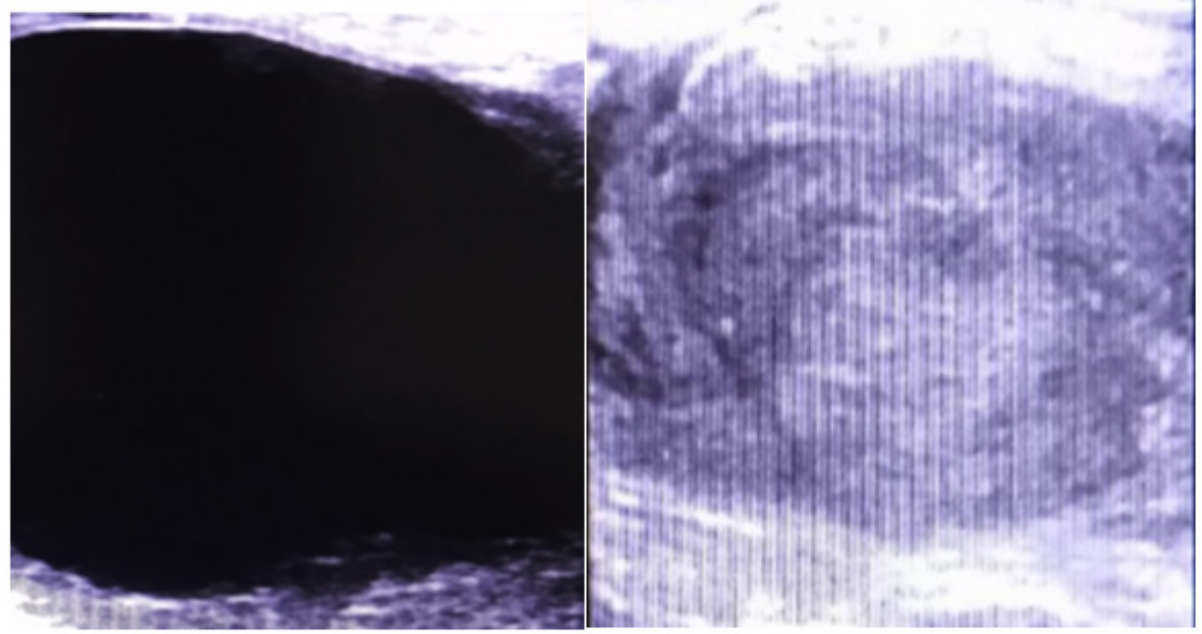
However, despite all these procedures the patient kept complaining of pain and swelling in the affected arm. At this hospital on examination the left arm was tense and swollen however non-tender; mild degree of swelling was noted in shoulder girdle and left chest wall as well. A doppler for deep venous thrombosis (DVT) came out negative however the B mode USG reported diffuse anechoic soft tissue collection in the

subcutaneous and deep fascia in left anterior chest wall, shoulder region and arm with small poorly defined irregular marginated hypoechoic soft tissues lesions broadly based on muscular chest wall. A percutaneous aspiration of these contents showed the contents to be serous in appearance. A repeat radiograph showed post bone grafting status of upper shaft of left humerus

with moth eaten appearance in rest of the medullary cavity and associated diffuse exuberant cortical thickening. The moth-eaten appearance in the spared medullary cavity added differential of a hematopoietic or lymphoreticular malignancy in addition to infection (Figures 1 & 2).



**Figure 1:** Radiograph left shoulder (AP View) shows an irregularly marginated large osteolytic lesion in proximal humeral shaft with exuberant cortical thickening..



**Figure 2:** B mode USG of left chest wall shows anechoic subcutaneous fluid collection with hypoechoic soft tissue deposits broad based of the muscular chest wall.



**Figure 3:** Chest CT shows a thick-walled cavity in superior segment of RLL.

The progressive pattern of disease made us to think for an alternative diagnosis. Thus, a decision to perform a screening whole body skeletal survey was taken which reported no other lesion. A detailed contrast enhanced computed tomography (CECT) of thorax and abdomen was done next day which revealed a thick-walled cavity in superior segment of right lower lobe and necrotic retroperitoneal lymph nodes, maximum measuring 17mm (max small axis diameter). After corroborating the radiographic findings of a permeative bone process in marrow of left humerus, a cavity in right lung and enlarged necrotic retroperitoneal lymph nodes, a possibility of tuberculosis and histoplasmosis was raised while reporting the CECT. A culture from the wall of lung cavity showed presence of *Pseudomonas aeruginosa* for which he was started on carbapenems. However, in view of negative cultures for tuberculosis and histoplasmosis, a further search for the exact etiology was continued. The haematological blood work-up showed a haemoglobin of 7.9gm%, TLC count of 12.2cells/mm<sup>3</sup>, DLC showing increased neutrophilic count, platelet count of 4 lakh cells/mm<sup>3</sup>. The LDH and S. alkaline phosphatase was raised 296 and 309U/L. The Serum calcium and inorganic phosphates were normal (Figure 3).

A neck ultrasound was performed next day which revealed few enlarged necrotic cervical lymph nodes. A decision to perform histopathological evaluation (HPE) of these nodes after excisional biopsy was taken. HPE picture showed obliteration of normal nodal architecture with closely packed small lymphoid follicles with non-cleaved cells and with moderate cytoplasm and multiple

nucleoli and suggestive of follicular subtype of lymphoma, stage II. With this surprising diagnosis six cycles of R-CHOP (rituximab, cyclophosphamide, doxorubicin hydrochloride, vincristine sulphate and prednisolone) regimen instituted however we lost the patient after two weeks because of fulminant opportunistic bacterial pneumonia due to immune-suppression.

### Discussion

Primary bone lymphoma is a rare variety of lymphoma and overall contributes to <1% of all cases. The most common subtype is diffuse large B cell lymphoma whereas follicular, Burkitt's, marginal zone lymphomas are rare or less common subtypes. It is mostly noted in young and mid aged adults in the age group of 20 to 50 years. The commonest presenting complaint is bone pains. Follicular lymphoma is an indolent tumor. The median survival period in most of the patients is about 12 to 15 years, although 10-15% may have an aggressive disease with a short survival period [6]. The radiological findings are usually non-specific and most of them show permeative bone destruction suggesting an infiltrative process which in most of the likelihood suggests a haematological malignancy however can still be confused with chronic or acute on chronic osteomyelitis type picture.

The mainstay of diagnosing follicular lymphoma is a detailed HPE work-up with immunophenotyping. The classic HPE picture is obliteration of normal nodal architecture with loss of

interfollicular spaces [7]. T (14;18) (q32: q21) is the hallmark chromosomal translocation noted in patients with follicular lymphoma. Immunophenotyping shows positivity for CD10, CD19, CD20 & CD22 cells. Clinico-radiologically a progressive pattern of disease on imaging and persistence, aggravation or suboptimal alleviation of signs and symptoms despite adequate treatment for osteomyelitis is a clincher for an alternative diagnosis. It has been suggested in the existing literature that an open bone biopsy be performed in all such cases where despite adequately treating the patient for osteomyelitis, the results remain un-equivocal.

## Conclusion

Only few cases of primary bone lymphoma's have been reported in existing literature and most of them have been misdiagnosed as osteomyelitis because of the rarity of this condition. A high index of suspicion of hematopoietic or a lymphoreticular malignancy should be kept in cases with equivocal response for osteomyelitis as the radiographic picture and clinical assessment may show considerable overlap. Follicular lymphoma mostly presents with nodal pattern; however, it may present primarily as an extra-nodal disease, and this should always be kept in mind.

## Declarations

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## Conflict of Interest

There were no conflicts of interest

## Ethics statement

Ethical approval was not required for this manuscript.

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