Asian Oncology Research Journal

4(1): 1-6, 2021; Article no.AORJ.64549

Diagnosis of Lytic Lesion in Proximal Tibia: Histiocytic Sarcoma, a Case Report

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Authors' contributions

This work was carried out in collaboration among all authors. Author CD designed the study, wrote the protocol and wrote the first draft of the manuscript. Authors GN and AP managed the analyses of the study. The work was carried out under the guidance of author AK. All authors read and approved the final manuscript.

Article Information

Editor(s):
(1) Sri Lakshmi Hyndavi Yeruva, Penn State Health, Hershey Medical Center, USA.
Reviewers:
(1) Victor Alves de Oliveira, Federal University of Piauí, Brazil.
(2) Walter Belda Junior, University of Sao Paulo, Brazil.
Complete Peer review History: http://www.sdiarticle4.com/review-history/64549

Case Report

Received 05 November 2020
Accepted 11 January 2021
Published 01 February 2021

ABSTRACT

Introduction: Histiocytic Sarcoma is a rare neoplasm which causes less than 1 percent of all malignancies of hematologic origin. The biological behavior of this neoplasm resembles that of lymphoma, however its cell line is histiocytic and anon-lymphoid. Thereafter, the malignant cell of the histiocytic sarcoma shows morphological and immune-phenotypic characteristics of the histiocyte of mature origin. In the most recent 2016 revision, the WHO classified histiocytic sarcoma inside the macrophage along with other histiocytosis and stroma derived dendritic cell tumors.
Case Report: A 70 year old female with Histiocytic sarcoma of the left proximal tibia which presented as a solitary lytic lesion with pain in the left knee since 3 months. We here in describe a rare differential to a solitary lytic bone lesion without constitutional symptoms which was treated by our own conventional method.
Conclusion: Though Histiocytic Sarcoma is a rare bony lesion, we should be aware of its presence and perform biopsy and immunohistochemistry in all lytic bone lesions.

Keywords: Histiocytic sarcoma; lytic lesion; proximal tibia.

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1. INTRODUCTION

Histiocytic Sarcoma is a rare neoplasm which causes less than 1 percent of all malignancies of hematologic origin [1]. The biological behaviour of this neoplasm resembles that of lymphoma, however its cell line is histiocytic and anon-lymphoid. Thereafter, the malignant cell of the histiocytic sarcoma shows morphological and immune-phenotypic characteristics of the histiocyte of mature origin. In the most recent 2016 revision, the WHO classified histiocytic sarcoma inside the macrophage along with other histiocytosis and stroma derived dendritic cell tumors [2,3].

The malignant cells of histiocytic sarcoma show positivity for one or more histiocyte lineage markers in immuno-histochemical analysis which includes CD68 (KP1, PGM1), CD163 and lysozyme. The markers which are often positive include CD45, CD45RO and HLA-DR. On the flip side, it is typically negative for Langerhans cell markers and other dendritic cell markers such as CD1a, langherin, CD21 and CD35 as well as for myeloid, B-lymphoid and T-lymphoid markers. S-100 protein may be positive but usually it expression is weak or focal and Ki67 has a variable expression [4].

Studies about the gene expression have shown that a subset of Histiocytic Sarcoma has a rearrangement of clonal type of the immunoglobulin gene, especially in cases associated with low grade B-cell lymphomas [5]. In some cases there has been description of the BRAFV600E mutation. A recent study conducted showed that 5 of 8 cases of Histiocytic Sarcoma presented the BRAF V600E mutation, making this a promising option for target therapy [6]. Also, recurrent mutations involving which involves the KM2D gene and MAP kinase pathway have been described [7,8,9]. Despite all the progress made with regards to Histiocytic Sarcoma, its etiology still remains to be known.

2. CASE REPORT

A 70 year old female presented with dull aching pain around the knee since 3 months which aggravated on walking following a fall at home. Examination around the left knee was normal without redness, swelling, deformity or sinus. On palpation there was no local rise of temperature, however slight tenderness was elicited on the medial tibial condyle. No crepitus or ligament laxity was demonstrated. The radiographs of the left knee showed an oval lytic lesion in the proximal medial tibial condyle with associated thinning of the cortex and no cortical breaks (Fig. 1).

The MRI of the left knee demonstrated a well defined, lytic, eccentric non expansile lesion involving the epi-metaphysis of medial tibia condyle without cortical break suggestive of Giant Cell Tumour/ Chondromyxoid Fibroma/ Metastasis (Fig. 2).

FDG -18 PET Scan revealed an FDG avid lytic-sclerotic lesion in the left medial and lateral tibial condyles with a metabolically active lesion in the medial tibial condyle with a few FDG avid cervical, retroperitoneal and mesenteric nodes, the largest measuring 1.03x0.55cm (Fig. 3).

High resolution Chest CT Scan, Blood and Serum investigations were normal. Based on the above findings, diagnosis of Giant Cell Tumour was considered, for which an excisional biopsy was planned.

A Standard Antero-medial approach was taken on the medial tibial condyle to explore the lesion. A window was made after confirming the tumour position under C-Arm. Intra-operative findings included the presence of a grey, fleshy growth which was completely removed. It was followed by curetting the cavity with a cylindrical burr, chemical cauterization was done with phenol and was filled with bone cement using the standard sandwich technique after confirming that the subchondral bone was not breached. Stability was provided by placing a L- shaped 7 hole plate over the medial aspect of the proximal tibia (Fig. 4).

Histopathological examination revealed Histiocytic Sarcoma of bone, confirmed by Immuno-histochemisty showing positive CD68, HLA DR, Lysozyme, EMA, Vimentin, S100 and negative CD38, 99, 23, 21, Desmin, P40 (Fig. 5).

Patient was treated according to the histopathology report by radiation to the left tibia, abdomen and chest as the tumour is sensitive to radiation. Physiotherapy was started on second post-operative day with range of movement for knee and toe touch being allowed. At 6 months follow up, the patient had no radiological and clinical signs of recurrence with full range of knee movement and with full painless weight bearing.

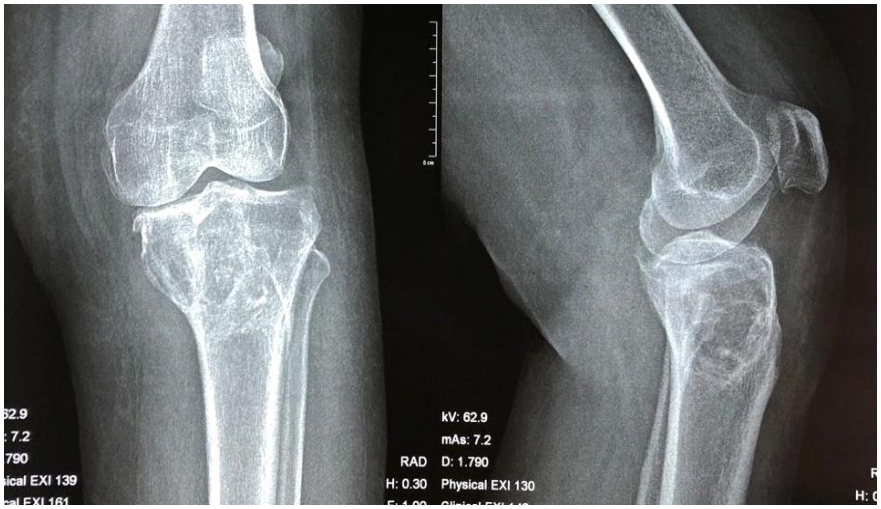

Fig. 1. Antero-posterior and lateral views of Left Proximal tibia showing lytic lesion in the Medial condyle

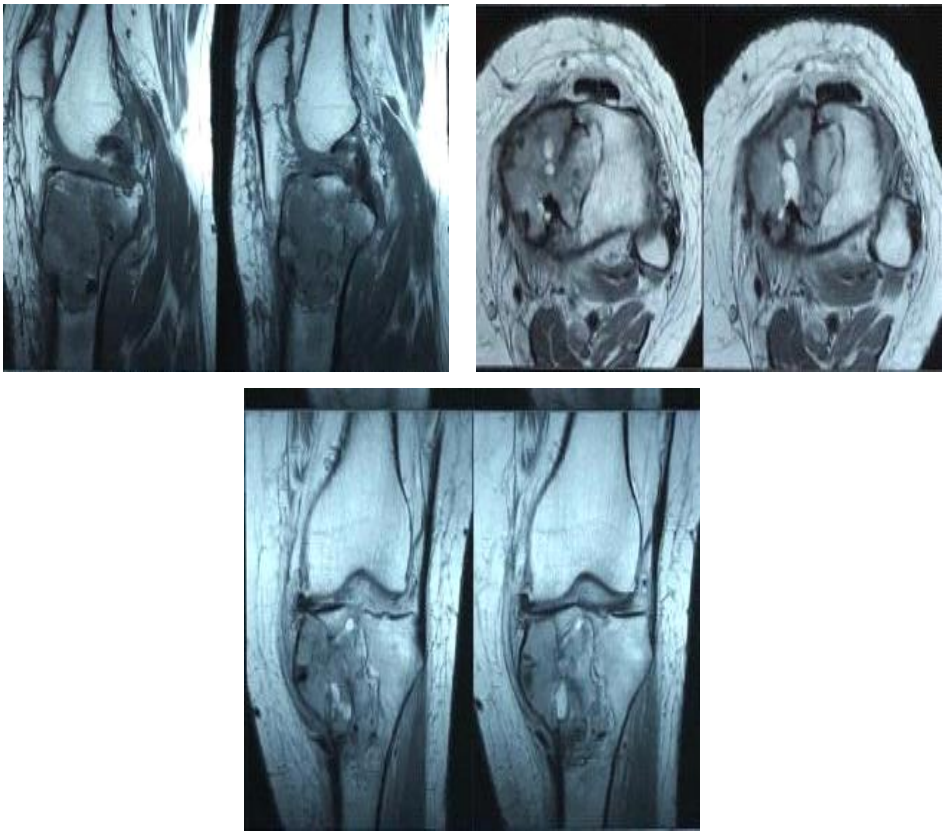

Fig. 2. Sagittal, Coronal and Axial slices of MRI showing the well-defined, lytic, expansile lesion in the left proximal tibia involving the medial condyle without cortical break

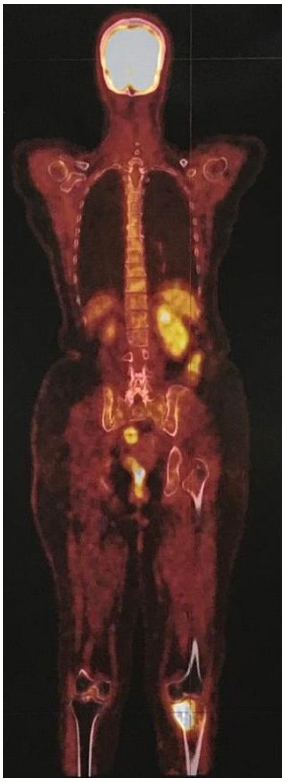

Fig. 3. PET scan image showing a metabolically active lesion in the left proximal tibia with a few FDG avid lymph node

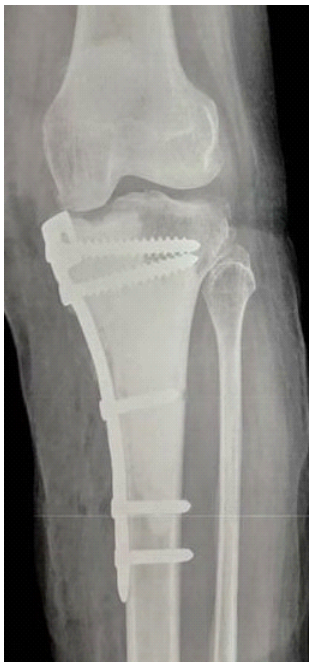

Fig. 4. Post Operative X ray showing wide local excision curettage of bony lesion with fixation with anatomical medial tibial plate using "sandwich technique" to fill the bony defect

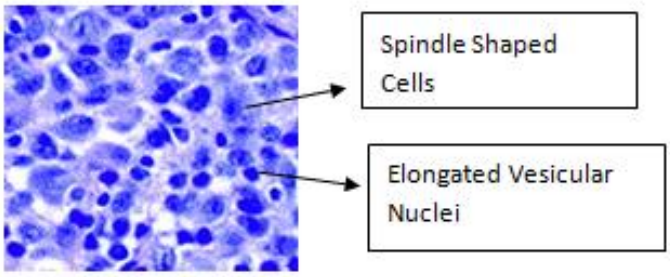

Fig. 5A. Reveal tumour composed of sheets of spindle shaped cells with elongated vesicular nuclei resembling histiocytes

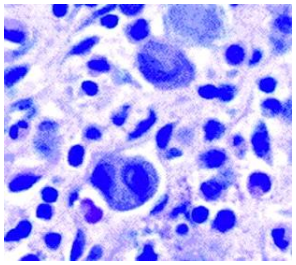

Fig. 5B. Reveal tumour giant cells with hyperchromatic pleomorphic nuclei

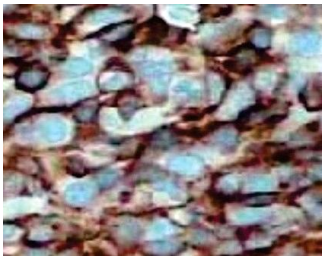

Fig. 5C. Shows CD 68 positivity of these cells

3. DISCUSSION

Histiocytic sarcoma is a rare and aggressive neoplastic proliferation of mature histiocytes [1,2] which is more commonly seen in adult males. Histiocytes are non-circulating cells from the bone marrow and are tissue based equivalents of monocytes expressing CD68 and CD163 (4,5). It is a true haematopoietic tumour and accounts for less than 1% of all haemato-lymphoid disorders . It is most commonly seen in lymph nodes and can present as painless masses in extra nodal sites like skin, liver, spleen, bone marrow and intestinal tract. It rarely affects the bone with only 4 cases reported in human beings (According to results shown by Pubmed when searched with the words “histiocytic sarcoma” and “bone” with the boolian operator “AND”) and no case reports for tibial involvement were found. The radiographic findings of X ray show lytic bone lesions [4,9]. This finding mimics Orthopaedic differentials like fibrous dysplasia, Giant cell tumour, Non Ossifying Fibroma, Osteoblastoma, Multiple Myeloma, Aneurysmal Bone Cyst and Hyperparathyroidism. MRI/ CT Scan cannot differentiate these bony lesions from the differentials mentioned above.

Histiocytic sarcoma is frequently accompanied by constitutional symptoms like weight loss, fever, etc with extra nodal involvement though in our case, there were no constitutional symptoms or weight loss. The patient presented with dull aching bony pain following a fall with a lytic lesion in the proximal tibia on X-Ray and MRI. The PET scan revealed no other metabolically

active lesions though a few lymph nodes, with the largest measuring 1.03x0.55cm were noted in the abdomen, but were reported non-conclusive. On excisional biopsy, after curettage, histopathology revealed Histiocytic Sarcoma which was considered as the last diagnosis because of its rarity.

4. CONCLUSION

Though Histiocytic Sarcoma is a rare bony lesion, we should be aware of its presence and perform biopsy and immunehistochemistry in all lytic bone lesions. The conclusion we can take from this case is that any Proximal Tibia Lesion should be suspected for Histiocytic Sarcoma although Giant Cell Tumour is more common.

CONSENT

As per international standard or university standard, patient's written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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Peer-review history:
The peer review history for this paper can be accessed here:
http://www.sdiarticle4.com/review-history/64549