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Case Report

Pathology Section

Plasma Cell Osteomyelitis: A Rare Entity

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ABSTRACT

Plasma cell osteomyelitis, characterised by abundance of plasma cells in the lesional bone is a rare entity. We report a case of chronic plasma cell osteomyelitis in a 14-year-old male child who presented with complaints of pain in right leg and fever. The X-ray AP view of right leg revealed a cavitary lytic lesion with a sclerotic border in proximal one third of right tibia. A clinical diagnosis of Brodie's abscess was favoured. Histopathological examination revealed sheets of plasma cells, many neutrophils and lymphocytes, areas of fibrosis and spicules of dead bone.

Immunohistochemistry revealed polyclonal population of plasma cells which were both Kappa and Lambda light chain positive. We recommend diligent histopathological examination and correlation with clinico-radiological profile for a correct diagnosis and in order to avoid an over diagnosis of neoplastic plasma cell lesion.

Keywords: Bone, Plasma cell dyscrasia, Plasma cell rich lesion, Sclerotic lesion

CASE REPORT

A 14-year-old male child patient presented to Orthopedics Out Patient Department of AIIMS Rishikesh, Uttarakhand, India, with chief complaint of pain in right leg and low grade fever (100° F) from last ten months. There was no swelling associated. The local examination revealed localised tenderness over anteromedial surface of proximal one third of tibia. The X-ray AP view of right tibia was suggestive of a cavitary lytic lesion with a sclerotic border in the proximal one-third of right tibia [Table/Fig-1a]. Baseline blood investigation revealed total leucocyte count and ESR to be within normal limits which indicates localise disease.

Based on above finding a clinical diagnosis of Brodie's abscess was suggested without any other differential diagnosis and curettage of the lesion was done and specimen was sent for histopathological examination.

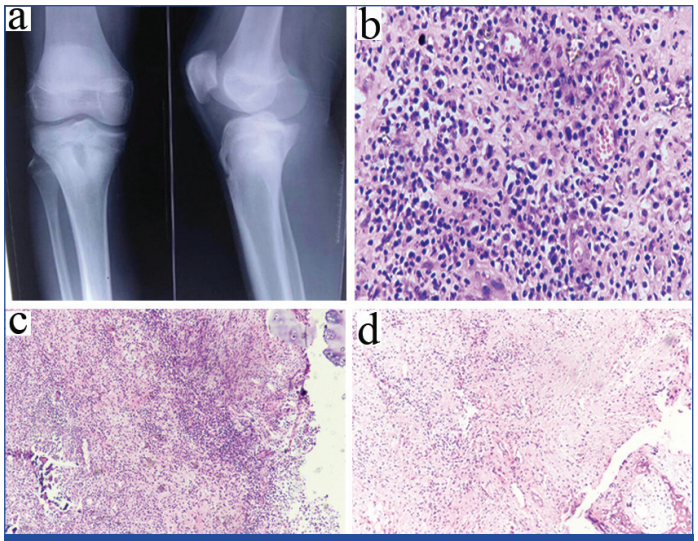
Gross specimen examination revealed multiple tissue pieces comprising of soft tissue as well as bony tissue aggregating to size 1×0.5×0.5 cm which were received and proper decalcification was done using 5% nitric acid in distilled water prior to routine processing.

On light microscopic examination, Haematoxylin and Eosin (H&E) stained sections showed thin bony trabeculae along with dense infiltration by mixed inflammatory cells comprising of predominantly sheets of plasma cells, many neutrophils and lymphocytes. Stromal oedema, many thick and thin walled vessels, areas of fibrosis and spicules of dead bone characterise by deep blue appearance were also noted [Table/Fig-1b-d]. Based on H&E, a differential diagnosis of plasma cell rich lesion including plasma cell dyscrasia, Lympho proliferative disorder and plasma cell osteomyelitis was made.

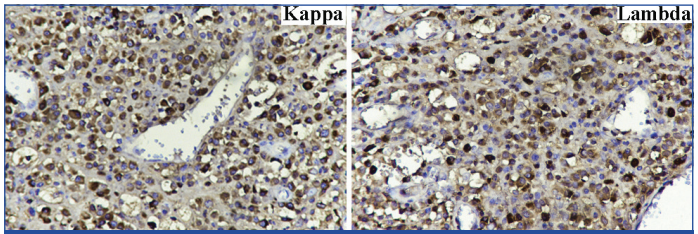
To differentiate reactive plasma cell from neoplastic plasma cell, immunohistochemistry for CD138, Kappa and Lambda was performed to look for clonality which showed positivity for CD138 and both light chain immune markers suggesting a plasma cell proliferation of polyclonal in nature [Table/Fig-2]. A final diagnosis of Plasma cell osteomyelitis was offered.

Curetted material from cavitory lesion of bone was also sent to microbiology lab for culture, which revealed occasional colonies of *Staphylococcus aureus*.

Patient was prescribed Non Steroidal anti inflammatory Drugs and after that patient did not come back for follow-up.



[Table/Fig-1]: a) The X-ray AP view of right leg revealed a cavitory lytic lesion with a sclerotic border in proximal one third of right tibia. Varied histomorphology of plasma cell osteomyelitis; b) Sheets of plasma cells, many neutrophils and lymphocytes (H&E, X400); c) Areas of fibrosis and spicules of dead bone (H&E, X100); d) Sheets of plasma cells and thin walled vessels (H&E, X100).



[Table/Fig-2]: Immunohistochemistry revealed polyclonal population of plasma cells which were both Kappa and Lambda light chain positive (Immunoperoxidase, X400).

DISCUSSION

Plasma cell osteomyelitis is a chronic recurrent, unifocal or multifocal inflammatory disorder, which has an unknown etiology. The clinical and radiographic features may make it impossible to distinguish from other sclerotic lesions of bone [1]. It is imperative that early and definitive diagnosis of the specific type of osteomyelitis is done to facilitate early and apt management of these patients. Owing to the rarity of plasma cell osteomyelitis, a clinical diagnosis is usually not considered and it is only on histopathological examination

that a definitive diagnosis is given. The infectious focus or foci may occur in different sites of the skeleton [2,3]. Plasma cell osteomyelitis usually attacks children and young adults, the most frequent site being metaphysis of a long bone [4]. In the present case, it was a 14-year-old male child with lesion in the right tibia. The pathogenesis of the plasma cell osteomyelitis possibly involves an abnormal sterile and aseptic immune process which is a response generated after infection. However, clinically may not show frank symptom of infection and hence is considered to be clinically silent [3]. The spectrum of histopathological changes can range from acute (acute inflammatory cell infiltrate, active bone resorption and necrosis, reactive bone formation) to subacute (predominantly lymphocytic and plasma cell infiltration) or chronic inflammation (fibroblastic organisation and bony sclerosis). Osteomyelitis is defined as an inflammatory process involving cortical and cancellous bone. It has a clinically undecided onset and with three distinctive zones on microscopy. The name plasma cell osteomyelitis is designated due to its characteristic histological findings, in which infiltration of plasma cells are seen. Histology comprises of a central zone localised in the center most area with dense infiltration of plasma cells, intermediate zone displaying fiber-rich scar tissue surrounding central zone and peripheral zone show fibrous and oedematous bone marrow with a protein-rich fluid [5]. In our case, biopsy showed numerous plasma cells, polymorphs and lymphocytes along with stromal oedema and many thick and thin walled vessels.

For reasons unknown, plasma cell osteomyelitis is noted in individuals having a strong host immunity and infected with organisms of low virulence.

It is noteworthy that the treatment of plasma cell osteomyelitis involves administration of Non Steroidal Anti-Inflammatory Drugs and antibiotics have limited role. This rare case presenting with non specific symptoms and clinical suspicion of acute osteomyelitis emphasises the need of careful work up. A histopathological examination is essential to establish the diagnosis and treatment should be started after confirmation of the histopathology of the lesion.

CONCLUSION

We recommend diligent histopathological examination and correlation with clinico-radiological profile for a correct diagnosis and in order to avoid an over diagnosis of neoplastic plasma cell lesion.

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