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Case Report

A rare case of plasma cell osteomyelitis with atypical presentation

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ABSTRACT

Plasma cell osteomyelitis is an uncommon type of localized chronic osteomyelitis which is characterized by the absence of typical clinical features of infection. It usually affects the metaphysis of long bones and presents as dense thickening of bone without sequestrum formation. We present an atypical case of plasma cell osteomyelitis with a lesion in the diaphysis of long bone and sequestrum formation. A 23 year old male patient came to our OPD with complaint of pain in mid-thigh radiating to the left knee since 2 months. X-ray showed radiolucencies in the medulla of middle third of shaft of femur along with a linear sequestrum suggestive of chronic osteomyelitis. An open biopsy was performed and surgical curettage was done. Histopathological examination was consistent with plasma cell osteomyelitis. Chronic plasma cell osteomyelitis is a rare condition with nonspecific findings and an unclear cause. Histopathology is the definitive tool for obtaining a diagnosis. Knowledge of this condition and proper management can give a better outcome, at the same time avoiding unnecessary investigations and procedures.

Keywords: Osteomyelitis, Plasmacytoma, Plasma cells, Sclerosis, Diaphyses

INTRODUCTION

Plasma cell osteomyelitis is a type of localized chronic osteomyelitis.[1] Primarly chronic osteomyelitis was classified as Brodie's abscess, plasma cell osteomyelitis, and sclerosing osteomyelitis (Garre) by Lennert (German literature) (1964).[1] Classification of primary chronic osteomyelitis varies in different parts of the world. In English speaking countries, classification is based on radiological criteria, whereas in German-speaking countries, it depends on the histological findings.[2] Exner and Cserhati, believed that each type of chronic osteomyelitis was a true disease entity.[2] Good literature is available on sclerotic osteomyelitis of Garre and Brodie's abscess but very little literature is published on plasma cell osteomyelitis in English literature. We discuss the presentation of plasma cell osteomyelitis and report a case of chronic primary plasma cell osteomyelitis with atypical features.

CASE REPORT

A 23 year old male patient came to our outpatient department with complaint of pain in mid-thigh radiating to the left knee since 2 months. Patient was apparently normal before that. The pain was dull aching in nature and aggravated during night and relieved with analgesics. There was no history of trauma, fever and constitutional symptoms. Physical examination showed mild tenderness in the mid-thigh region. There was no local rise of temperature, no scars, sinuses, swelling, erythema. Knee range of movement was full and painless. Radiographs showed irregular thickening of the cortex with sclerosis and radiolucencies in the medulla of middle third of shaft

of femur along with a linear sequestrum suggestive of chronic osteomyelitis (Figure 1).

Laboratory investigations showed total white blood cells (WBC) count 8,500/mm^3, with 60% neutrophils, 30% lymphocytes, 8% eosinophils, 2% monocytes and ESR-25 mm/h. Renal function tests, viral markers and coagulation profile were all in normal limits. An open biopsy was performed and surgical curettage was done through the lateral approach. Bone and bone marrow was sent for gram staining, histopathological examination and culture and sensitivity.

Gram staining showed gram positive cocci in clusters and few pus cells. Culture and sensitivity showed coagulase positive staphylococci, which is sensitive to cefotaxim, amikacin and ofloxacin. Histopathological examination showed irregular fragments of devitalized bone surrounded by dense fibrous tissue heavily infiltrated by uni- and bi-nucleated plasma cells, lymphocytes and few granulocytes along with extensive areas of haemorrhages, fibrin and few lamellar viable bony trabeculae with dilated sinusoids (Figure 2 and 3).

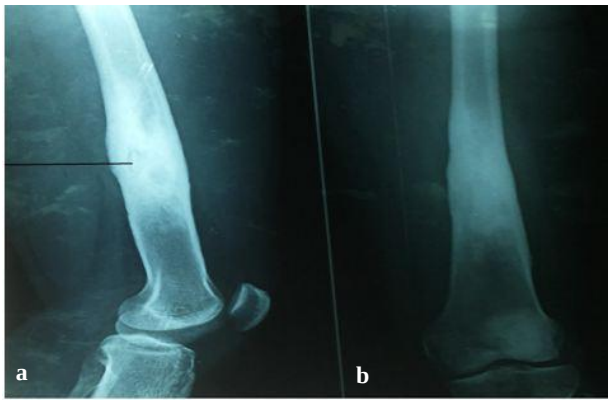

Figure 1: Antero-posterior and lateral radiograph of lower two-thirds of femur and knee joint of irregular thickening of the cortex and sclerosis in the middle third of shaft of femur.

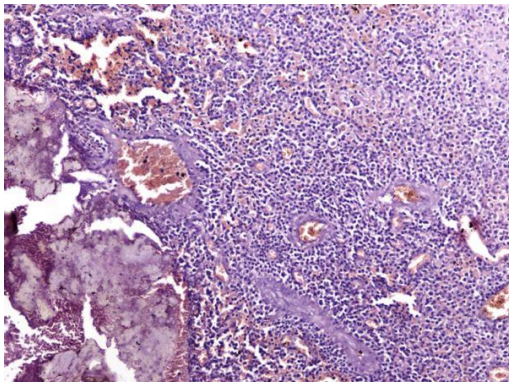

Figure 2: Histopathological examination of irregular fragments of devitalized bone surrounded by dense fibrous tissue heavily infiltrated by uni and binucleated plasmacells.

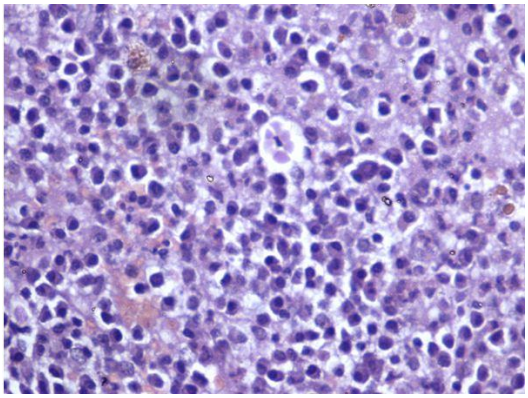

Figure 3: Magnified view of the specimen of Russel bodies (extra cytoplasmic immunoglobulins) extra cellular eosinophilic spherical bodies.

Findings were consistent with plasma cell osteomyelitis. The patient had an uneventful post-operative course. He was placed on oral ofloxacin for 4 weeks. Follow-up examination 3 months later, revealed complete resolution of symptoms and a complete recovery.

DISCUSSION

Plasma cell osteomyelitis is a primary chronic hematogenous osteomyelitis with clinically undecided onset.[1] It is thought to develop when body's resistance is good and virulence of organism is restricted. It is most commonly seen in patients between the age groups of 10 to 20 years and more between 21 to 40 years.[1] The male to female ratio is 3:2.[1] The clinical course is slow and inactive with occasional symptoms of common cold with fever, especially of tonsillitis or sinusitis.[1] The main symptom is recurrent pain, particularly at night. Rarely, may present with fever and effusion in the surrounding joint.[2] The patients are usually not emaciated and the local findings are unremarkable. The same was the presentation in our case without fever or joint involvement.

Plasma cell osteomyelitis and acute hemorrhagic osteomyelitis are similar in that the frequently involved sites are metaphyseal long bones. Plasma cell osteomyelitis and multiple or solitary myeloma are similar in that frequently involved bones are the vertebral body.[1] The most frequently involved bones in Plasma cell osteomyelitis were metaphysis of long bones, and also the lower thoracic and upper lumbar vertebral bodies. It is usually a solitary lesion, rarely polyostotic.[3] In our case, there is involvement of the diaphysis of femur.

Cserhati noted various findings; from coarseness to disappearance of structure, from indistinct radiolucency to polycystic appearance with durable sclerotic border. The findings varied by reason of diversity of pathological changes.[1] There were no specific radiological features which made it possible to differentiate this condition from other diseases such as bone tumours, with absolute

certainity.[2] Radiologically, there is lytic lesion surrounded by osteosclerotic border with no sequestrum.[3]

Our case differed from the classical presentation of plasma cell osteomyelitis in that there is sequestrum formation which gave us the probable diagnosis of chronic osteomyelitis. But because of the lesion in the diaphysis, which is not a classical location for osteomyelitis, the diagnosis of tumour could not be ruled out. Laboratory examination showed no abnormalities in many of the reported cases.[1-4] likewise in our case also immunologic studies revealed no abnormalities; this point is clinically different from solitary plasmacytoma.[1] In solitary myeloma there are a normal cell pattern of the sternal marrow, normal electrophoretic pattern of the plasma proteins and absence of Bence Jones protein in the urine.[1]

Only histological studies of the lesion make it possible to differentiate it from plasma cell osteomyelitis. Histopathologically, plasma cell osteomyelitis is described by the terms 'osteomyelitis albuminosa' (whitish mass) and 'osteomyelitis plasma cellulare' (consists almost entirely of plasma cells).[3]

Microscopically, Exner described distinctive with three zones which were composed of central, intermediate and peripheral zone marked infiltration of plasma cells with granulation tissue in the center of the inflammatory focus (central zone), proliferation of fibrous tissue around the center (intermediate zone) and fibrous bone marrow with protein-rich fluid in the periphery of the focus (peripheral zone).[1] In plasma cell myeloma (solitary plasmacytoma), the histological findings in the central and peripheral areas reveal a monotonous appearance, in which occasionally plasma cells are poorly differentiated with bizarre cells, and bone trabeculae show only process of destruction. Fibrosis and vascularization are not conspicuous, and plasma cells arrange adjacently with each other. It is sometimes, however, difficult to differentiate plasma cell osteomyelitis and plasma cell myeloma (solitary plasmacytoma) microscopically.[1]

Our pathologists couldn't make out the classical three zone description but the presence of irregular fragments of devitalized bone surrounded by dense fibrous tissue heavily infiltrated by uni- and bi-nucleated plasma cells prompted the diagnosis of plasma cell osteomyelitis. While the cause in many cases remains unclear, the leading theory seems to suggest an occult infection, however, on review of literature, cultures have been positive in only 8% of cases in lesions involving bones other than the mandible.[4] Sampling errors or the indolence of the process may contribute to the low incidence of culture positivity.[4] In the examination of the causative organism of plasma cell osteomyelitis, *Staphylococcus aureus* had been proved in many cases as was with our case.[1] This case of plasma cell osteomyelitis was atypical for reasons like involvement of the diaphysis, presence of sequestrum and culture positivity for *Staphylococcus aureus*.

Most of the cases described in literature were managed with curettage of the lesion followed by a complete course of culture specific antibiotics.[1-4] We also advocated curettage of the lesion at the time of biopsy, followed by a course of culture-specific antibiotics (ofloxacin) for 4 weeks. Few cases described in literature were mistaken for a malignant bone tumour and were treated with wide resection and bone grafting.[1]

CONCLUSION

In summary, chronic plasma cell osteomyelitis is a rare condition with nonspecific findings and an unclear cause. The clinical and radiographic features may make it impossible to distinguish from other sclerotic lesions of bone. Atypical features make this condition even more difficult to understand. Only histopathology is the definitive tool for obtaining a diagnosis. It shows the importance of biopsy before proceeding with definitive treatment. Knowledge of this condition and proper management can give a better outcome, at the same time avoiding unnecessary investigations and procedures.

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