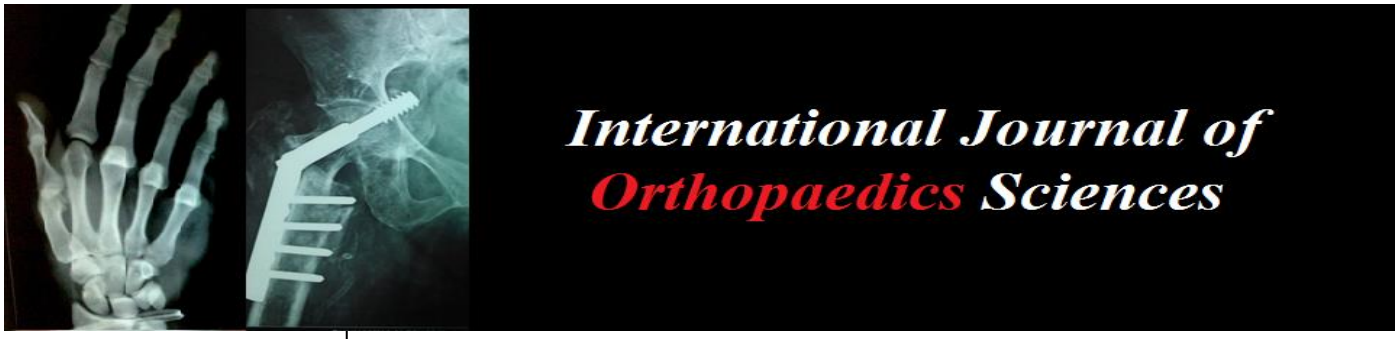

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Plasma cell osteomyelitis with atypical growth, in open fracture both bone leg-a rare entity

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Abstract
Plasma cell osteomyelitis (PCO) is a rare type of chronic osteomyelitis usually affecting metaphysis of long bones and characterized by an abundance of plasma cells in the lesion with a distinct histopathological pattern. It has been reported in various age groups, more commonly in the second to fourth decade of life. It is usually primary in origin and has been mostly reported in non-traumatic cases. It is characterised by a quiescent inflammation with abundance of plasma cells in the lesional bone. We are hereby reporting a case of chronic secondary post traumatic plasma cell osteomyelitis, in a 52yr male adult who reported 20 months post multiple surgeries on an open both bone leg fracture, presenting with exposed avascular bone over proximal one-third leg. Atypical organisms were reported on cultures and patient was started on appropriate antibiotics and reported sterile after 8 months of therapy. Patient underwent bone resection and reconstructive procedures for limb salvage.

Keywords: Plasma cell, post traumatic, atypical organisms, periosteal reaction, hypoxia

1. Introduction
Plasma cell osteomyelitis is a rare entity of primary chronic non-suppurative osteomyelitis (PCNSO), seen secondary to an abnormal sterile and aseptic immune response generated after infection [1], where pathological studies of the specimen show zonal differentiations with central zone containing sheets of plasma cells [2-5]. It's a rare pathology with a diagnosis that is not easy to establish clinico-radiologically [1-5]. This type of osteomyelitis is mostly commonly found in metaphysis of long bones [1-3] as in our case study but cases of atypical presentations in spine[4], diaphysis of long bones and phalanges have also been found. It is commonly seen in the second to fourth decade of life although cases in other age groups have also been reported. The male to female ratio is 3:2 [3]. It has been found incidentally in patients who have a strong immune response against the infectious microbe invading the bone [1-3]. Clinical diagnosis is usually not considered and a high index of suspicion is required. Diagnosis is mainly confirmed by histopathological examination [1-4].
Plasma cell osteomyelitis, as a variant of primary chronic osteomyelitis, has been frequently confused with Garre's osteomyelitis, sclerosing forms of osteomyelitis and benign solitary neoplasms in view of its indistinct clinic-radiological picture [1-2]. PCO is most commonly associated with sclerosing forms of osteomyelitis on radiology, the only difference being the absence of sequestrum and periosteal reaction [2] in PCO as compared to other forms of osteomyelitis. On histopathology, it can be divided into 3 zones, i.e, and central zone of granulation tissue in the center of the inflammatory focus densely infiltrated with plasma cells, intermediate zone of fibrous scar tissue and peripheral zone of fibrous bone marrow with surrounding edema [2-3]. Like all other Osteomyelitis, the most common organism isolated from this variant is Staphylococcus aureus [1-4] although it is assumed that infection can occur with any bacteria of low- virulence in a host with good immunity. In literature, plasma cell osteomyelitis has not been associated commonly with trauma [2-3] but here we are reporting a case of plasma cell osteomyelitis secondary to trauma, caused by atypical organisms and presenting with a sequestrum and periosteal reaction which was diagnosed on CT radiography.

Case report

A 52 year male reported to us with exposed avascular bone over proximal one-third tibia for last 6 months. Patient had a road traffic accident 2 years ago followed by open fracture Right Leg for which he underwent multiple surgeries at regular intervals. There was no history of fever or wound discharge. Consent for examination, investigation and subsequent management was taken from the patient. Clinical examination revealed two raw areas measuring 5x4cm and 4x3cm over right upper proximal tibia with exposed avascular bone (Fig 1). The movements at knee and ankle joints were restricted with the knee joint fixed in extension and the ankle fixed in equinus position. Conventional radiology of the proximal tibia showed an ill-defined bony lesion with heterogenous density with periosteal reaction (Fig 2). Subsequent CT scan to further define the lesion showed a comminuted fracture of tibia at the proximal end with multiple cortical bony defects, hyperdense fragments suggestive of sequestrum with surrounding periosteal reaction were noted.

Lab investigations revealed a mild leukocytosis while erythrocyte sedimentation rate and C-reactive protein were normal.

Operative Steps

Under combined spinal epidural anesthesia, the patient was taken up for debridement and sequestrectomy. Necrotic bone (12X4 cm) was removed from the proximal part of tibia, underlying bony bed was debrided uptil underlying bleeding bone. Debridement and pulse lavage with 10 litre Normal Saline was done. Following debridement and removal of nacrotic tissue from proximal tibia bone gap was created which was bridged with an antibiotic PMMA spacer. Proximal soft tissue defect anteriorly was covered with a fasciocutaneous flap in proximal tibia. Limb was stabilized using a spanning External fixator and nacrotic bone submitted for microbiology and culture (Fig 3).

On Histopathological examination of the bone specimen, nacrotic bony tissue was noted, surrounded by fibrous infiltrate and granulation tissue which included predominantly plasma cells, few lymphocytes and histiocytes. Fibrous tissue with plasma cell infiltrates were also found in the perivascular region in the inflammatory zone (Fig 4, 5, 6). No evidence of neoplastic aetiology was found. Culture and sensitivity of tissue revealed atypical organisms such as Proteus, Klebsiella and E. coli and antibiotics with sensitivity to the same (IV Cephalosporins 2nd generation) were started and continued for 3weeks.

Proximal antibiotic spacer was removed after 3 weeks and corticotomy in distal tibial metaphyseal fragment was done following which distraction histiogenesis (1mm/day) using a rail road fixator was started, and continued for over a period of one year. Patient is now being followed up to note the progress of the limb lengthening procedure (Fig 7). No fever, recurrence of infection in proximal tibia has been noted at one year follow up.

Discussion

Very few studies are available regarding Plasma cell osteomyelitis (PCO) in orthopaedic conditions e.g. metaphyseal regions of long bones, vertebrae and foot pathology [1-4] Usually it has been associated with non-traumatic conditions [2-3] while PCO in traumatic ortthopedic conditions has not been reported so far.

Plasma Cell Osteomyelitis commonly affects young adults between the second to fourth decade3 while some reports have come up with a bimodal distribution 1. The present patient reported in fourth decade to us.

PCO usually presents with a quiescent inflammation of the bone associated with abundance of plasma cell infiltration with sclerotic proliferation of fibrous tissue without suppuration [1-2]. A similar presentation has been noted in our case study as well where the patient presented with complaints of exposed avascular bone without any associated systemic symptoms or discharge from wound site.

Radiology in patients with PCO presents as solitary lesions which may be lytic or sclerotic with usually no evidence of surrounding periosteal reaction [2]. Presence of a sequestrum may not be present [1-2] in all cases although sequestrum formation and periosteal reaction was there in our patient.

While clinico radiological presentation of plasma cell osteomyelitis mimics that of solitary neoplasms and variants of sclerosing osteomyelitis, histopathological examination of tissue is of paramount importance to arrive at definitive diagnosis [1-3]

On histopathology [1, 3-4], a central zone of granulation tissue and plasma cells, with a surrounding intermediate zone of fibrous tissue and a peripheral zone of fibrous bone marrow and protein-rich fluid (Exner) is noted. Although the three zones could not be distinctively identified as per Exner's description in our case study, necrotic bony tissue surrounded by a proliferation of fibroblasts and granulation tissue with a abundance of plasma cells was noted.

For reasons unknown, plasma cell osteomyelitis is noted in individuals who have a strong host immunity [1-3]. Staphylococcous Aureus is commonly associated with PCO [2-3] unlike our case where atypical microorganisms e.g. Klebsiella, Pseudomonas, Proteus, E.coli were isolated. This could be due to anaerobic wound conditions which favour growth of Klebsiella and Pseudomonas. Anaerobic conditions favour plasmablasts and plasma cell formation5 with a suspected role in plasma cell tumorigenesis [5]. Both the anaerobic organisms and the infiltrating immune cells in the host consume oxygen contributing to hypoxia eventually lead to such infections. In this manner, hypoxia shapes the host–pathogen interaction [6].

Most of the cases described in literature were managed with curettage of the lesion [1-3] and NSAID therapy[3]. Due to identical radiological presentation PCO is likely to be confused with solitary myeloma [2-3] and wide resection has been erroneously done in such patients. Needle biopsy of suspect lesion followed by histopathology is mandatory to avoid pitfalls in diagnosis and management of such lesions. Culture sensitivity of isolated organisms followed by appropriate antibiotics for 8 months helps in healing of lesion. In present case, in view of the large surface area of the lesion (12cm), and the need for curettage and excision of the same resulting in significant shortening of the limb, bone transport using Rail road fixator was done to address resultant bone gap.

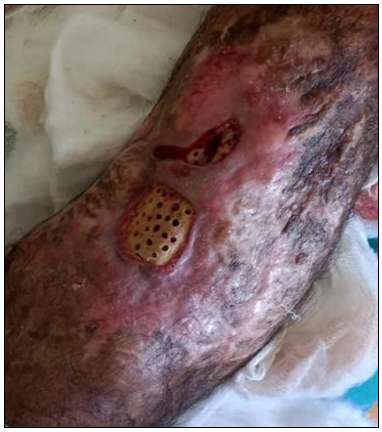

Fig 1: Two raw areas of 5x4 and 4x3cms over proximal tibia at presentation

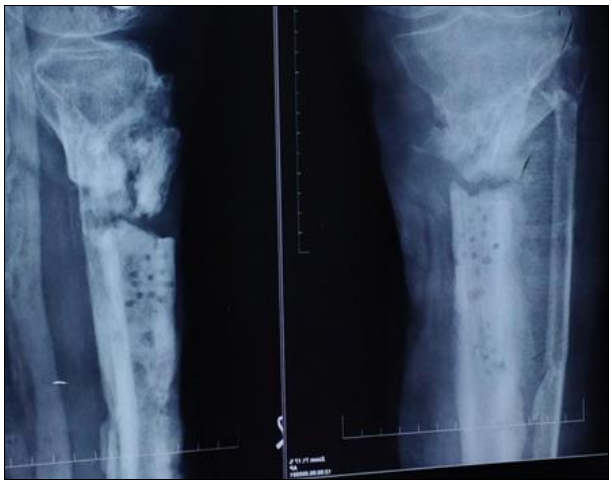

Fig 2: Conventional X-rays of proximal tibia at presentation, post a drill whole procedure with evident periosteal reaction around bone

Fig 3: Conventional X-rays of proximal leg after resection of sequestrum, post antibiotic spacer removal, stabilised by a rail road fixator

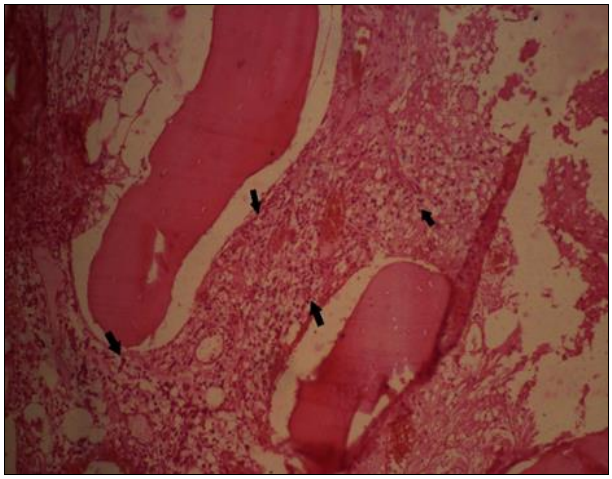

Fig 4: Lesion a 200x magnification showing diffuse infiltration of plasma cells (Arrow marks) with proliferation of capillaries and fibrous tissue

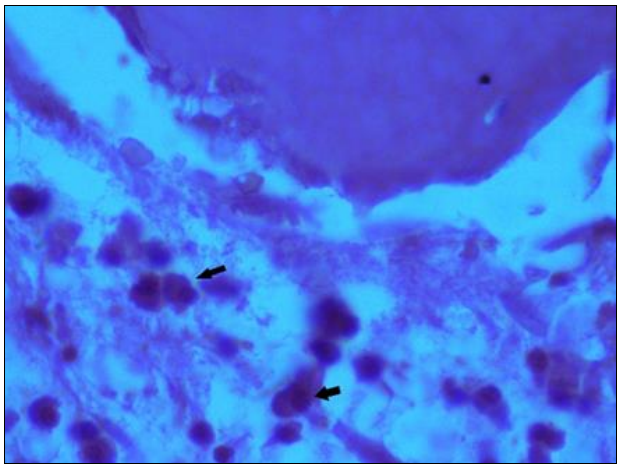

Fig 5: Lesion at 1000x magnification showing plasma cells with Russel bodies (arrows) examined under Wright-Giemsa stain

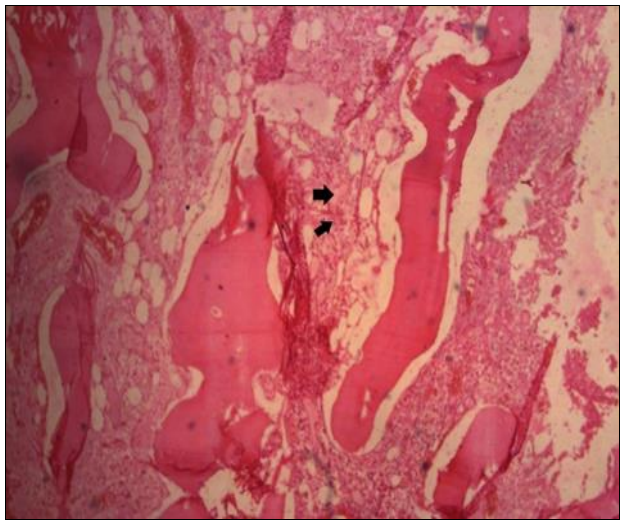

Fig 6: Lesion at 100x magnification showing the perivascular infiltration of plasma cells and fibroblasts

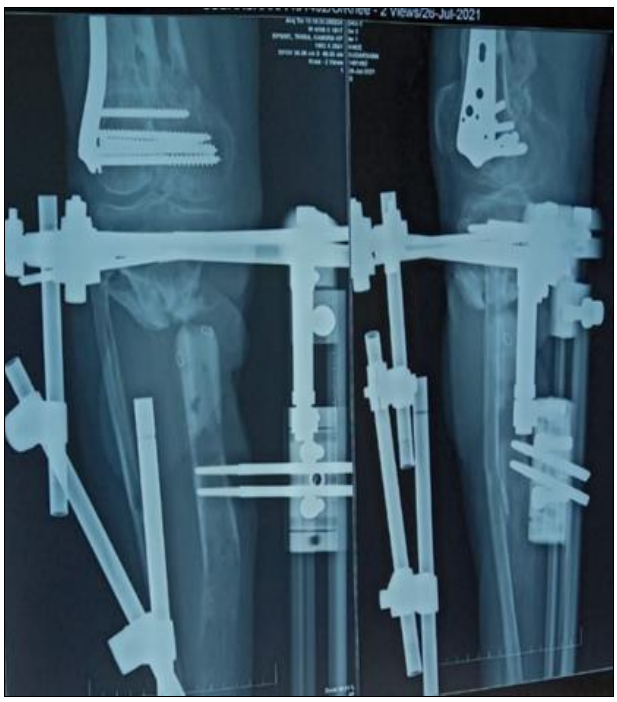

Fig 7: Conventional X-rays during the final stages of bone transport where the bone defect is almost filled

Conclusion

Chronic plasma cell osteomyelitis is a rare condition with nonspecific findings and an unclear cause. The clinic radiological similarity with solitary bone lesions makes histopathology an important step in establishment of appropriate diagnosis and initiation of proper treatment. Knowledge of this rare but manageable condition can go a long way in achieving better patient outcome.

Conflict of interest

The authors declare no conflict of interests.

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