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Sclerosing Osteomyelitis of Garre of Tibia Diaphysis Masquerading as Recalcitrant Medial Tibial Stress Syndrome

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Abstract: Chronic sclerosing oteomyelitis of Garré is an uncommon form of bone infection with variable clinical presentations. Symptomatic cases have a swollen, tender segment of the affected bone that may require surgical decompression in refractory situation. The diaphyseal origin is a rare site and the diagnosis requires exclusion and histopathological correlation. The present case cites this lesion in tibial diaphysis, a rare site. The case was managed as medial tibial stress syndrome, a common overuse injury in atheletic children, before appropriate diagnosis and treatment. The report highlights importance of anticipating some underlying pathology in refractory cases and assistance of advance imaging technologies to help diagnose and treat them.
Keywords: Osteomyelitis, Bone Infection, Tibia

INTRODUCTION

A non - suppurative, sclerosing form of bone infection causing distension of bone by cortical thickening was described by Garré [1]. The disease entails a tender, localised segment of involved bone [1]. The common sites are mandible and long bone metaphyses especially in pediatric age group [2]. Diaphyseal involvement is rare and can mimic other conditions requiring careful exclusion with appropriate use of clinical acumen and investigative battery. The fact that most of the times bacterial cultures are negative adds to complexity of diagnosis [3, 4]. The clinical and early radiological features of the disease are difficult to be distinguished from medial tibial stress syndrome in the settings of endurance sports. This relates to the fact that shin and the knee region bear the brunt of overuse symptoms in long distance runners [5].

CASE REPORT

A twelve year old child presented to us with history of chronic recalcitrant pain over the left shin at junction of middle and lower third for last nine months. He was an athlete and long distance runner. The pain started insidiously since last nine months, mild and intermittent at the beginning but increasing in magnitude and duration lately. There was no diurnal variation, radiation or referral pattern associated. Pain increased on prolonged weight bearing and relieved by rest but transiently. The affected part of the bone has got swelling that was apparent when compared to contra-lateral extremity. There was direct tenderness but no localized increase in temperature and overlying skin was not adherent to the affected region. The swelling was bony hard in consistency and appeared part of the bone. There was no other complaint remote or related to the present condition. There were no 'red flags' suggestive of acute infective process or neoplastic lesion. The patient was a healthy child with appropriate weight for his age and height as per the pediatric consult. The child was elsewhere managed as medial tibial stress syndrome before coming to us.

Relevant blood and ancillary investigations were unremarkable. The radiographs of the affected leg revealed diaphyseal circumferential cortical thickening over region corresponding to the middle and distal third junction of tibia with associated sclerosis (Fig.1). There was no nidus or lucent foci visible on plain radiographs. A provisional diagnosis,on the basis of radiological evaluation, was made of a subacute form of osteomyelitis like sclerosing osteomyelitis of Garre till proved otherwise. Differentials included chronic non-pyogenic infection, Brodie's abscess or osteoid osteoma. Magnetic resonance imaging (MRI) of the affected leg for additional information corresponded to the provisional diagnosis of chronic infective osteomyelitis with sclerotic cortical segment of bone ,distension and obliteration of medullary canal with sclerosed bony tissue. The unavailability of bone scan precluded this evaluation (Fig. 2).

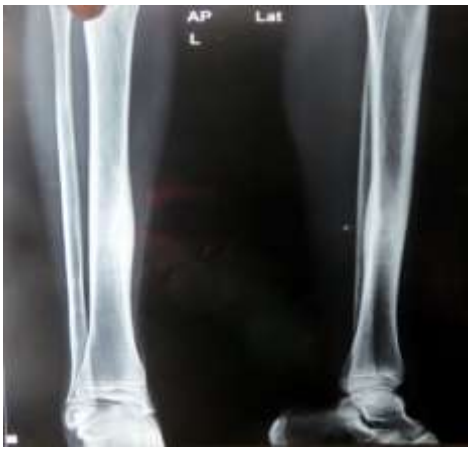

Fig. 1: Radiograph showing thickened, sclerosed and fusiform segment of affected bone

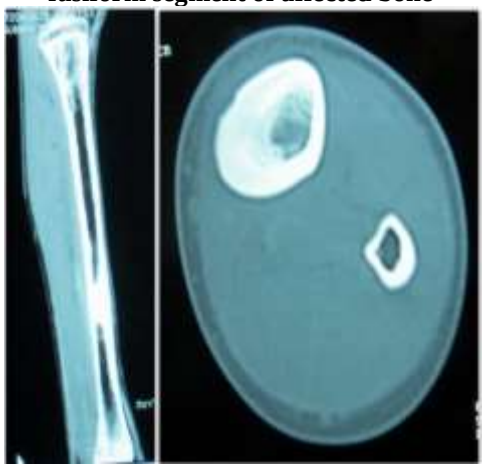

Fig. 2: MRI of the lesion to delineate pathology and extent

The patient was planned for surgical management in view of refractory trial of conservative treatment that included rest, pain medications, empirical broad spectrum antibiotic along with supportive, symptomatic management for two months. The surgery was done after proper written informed consent by parents in view of the case being minor. The tibia was accessed through antero-lateral approach with incision as per our requirement. A longitudinal cortical trough over the affected segment of the bone was fashioned using multiple drill-hole technique. A cortical de-roofing in this way was followed by localized intramedullary curettage. The sclerosed and abnormal bony tissue was removed from cavity till the endosteal bleeding was apparent and obstructed medullary cavity was recanalised with drilling on either side till healthy marrow from normal bone oozed out (Fig. 3). The curetted matter along with part of marginal healthy bone prepared for investigations. There was no facility of intra-operative frozen section by experienced pathologist to ascertain the limit of healthy and abnormal part of the bone, so we relied on clinical and gross macroscopic evaluation at the time of surgery.

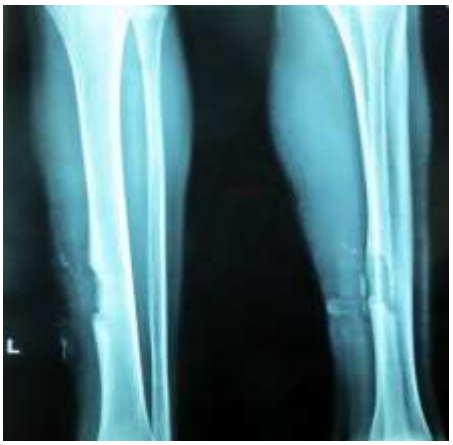

Fig. 3: Immediate post-operative radiograph showing cortical trough

The collected specimen had no evidence of purulent material. It was sent for Gram stain, Acid Fast stain and fungal culture along with culture sensitivity and histo-pathological analysis. The investigation for identification of causative organism was unremarkable and histopathological report underlined presence of chronic infective lesion affecting the bone.

RESULT

There was dramatic improvement in all clinical parameters including pain profile. Over a period of two weeks postoperatively, the pain gradually decreased to a state of no pain and the child resumed weight bearing to tolerance. The child was asymptomatic since 2 months postoperatively. The surgical wound healing was uneventful and there was no subsequent morbidity with respect to the cortical trough site during follow up till one year. The cortical defect gradually filled with normal bone in serial radiographic studies (Fig. 4). The child was asked to remain in periodic review in view of any complication or recurrence of the lesion.

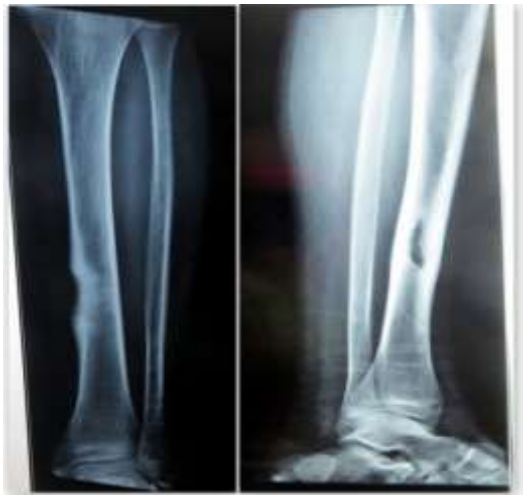

Fig. 4: Three month post operative radiograph showing healing bone defect

DISCUSSION

Medial tibial stress syndrome and stress fracture has been common overuse injury patterns in long distance runners [5]. Medial tibial stress syndrome is an "exercise induced localized pain along the distal two third of posterior medial tibia" [6]. Chronic recalcitrant pain not responding to conservative management require further investigations to rule out any sinister underlying pathology. Sclerosing osteomyelitis of Garré is a chronic form of infection with indolent course and varied pattern of symptoms between waxing and waning episodes. The disease has no exact etiology as recent work suggests certain genetic and autoimmune factors being at play [7, 8]. Mostly these cases are managed by conservative and supportive treatment [9]. A prolonged course of disease along with culture negative status and biopsy suggestive of chronic infective pathology are key features supporting the diagnosis of Garré osteomyelitis [9]. A cortical trough and opening of obliterated medullary cavity has been advocated for pain relief [10]. The disease has been related to other conditions affecting bone like chronic recurrent multifocal osteomyelitis(CRMO) and synovitis, acne, pustulosis, hyperostosis and osteitis (SAPHO)syndrome [9, 11].

CONCLUSION

The report highlights importance of anticipating some underlying pathology in refractory cases and assistance of advance imaging technologies to help diagnose and treat them. Symptomatic cases refractory to a trial of this form of management may benefit from surgical methods.

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