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Calcaneal osteosarcoma, a challenge for diagnosis: a rare case report and literature review

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Abstract

Osteosarcoma of calcaneum is of rare occurrence. Infrequent occurrence produce dilemma and challenge to it's diagnosis, leading to a delayed treatment. Here we present a rare case of chondroblastic variety of calcaneal osteosarcoma in an adolescent that produced a dilemma for diagnosis. Here also we have discussed the available literature regarding the clinical, radiological and histological features of calcaneal osteosarcoma.

Keywords: Calcaneum, osteosarcoma, chondroblastic type, dilemma in diagnosis, rare occurence

Introduction

Though osteosarcoma is the 2nd most common primary malignancy of bone, involvement of foot as a site is rare. Pedal osteosarcoma has an incidence rate of 0.2-2% [1]. Among the tarsals calcaneum is the favoured site [2]. Rarity of occurrence is the main reason behind it's missed and delayed diagnosis [2,3]. According to some authors osteosarcoma of foot constitute a different spectrum of osteosarcoma with slightly more favourable outcome [2,4,5]. The key to long term survival being early diagnosis and management, a high degree of suspicion is needed while evaluating a mass lesion of calcaneum. Here we present a rare variety (chondroblastic) of osteosarcoma of the calcaneum and showed how diagnostic confusion lead to delayed diagnosis of the lesion.

Case presentation

A 15 year hindu male presented to our outpatient department with complain of pain in the left heel and widened appearance since last 1 year. Pain was insidious onset, slowly progressive, dull aching, none radiating, aggrevated by activity and relieved with rest & analgesics. On examination there was circumferential swelling of the left calcaneum, bony hard in consistency, with an ill defined border & dilated tortuous vein on surface but no scar or sinus. There was no local rise of temperature and progression of growth being accelerated in last 3months (**Figures 1** and **2**). Patient had a bunch of x-rays and two inconclusive FNAC reports. X-ray showed a dense sclerotic lesion involving the left calcaneum, with abundant periosteal reaction creating a sunburst appearance and infiltrating the surrounding soft tissue (**Figure 3**). NCCT with 3D confirms the diffuse periosteal reaction of calcaneum (**Figures 4** and **5**). MRI showed heterogeneous mass of low intensity involving the entire calcaneum with infiltration and oedema of surrounding soft tissue. NCCT of chest showed no sign of metastasis. Other biochemical test showed no abnormality. Biopsy was taken with lateral incision and tissue sent for histopathology study. Histopathology section showed a malignant tumor infiltrating the bone with exuberant osteoid formation. Individual cell showed moderate cytoplasm, enlarged irregular hyper chromatic to vesicular nuclei with coarse chromatin with prominent nucleoli. Many bizarre nuclei with atypical mitosis were seen. Many areas showed destruction of bone and cartilage. Other areas showed chondroid degeneration (**Figure 6**). Patient was undergone a below knee amputation and has a no local recurrence at 1 year follow-up.

Discussion

Osteosarcoma of foot is of rare occurrence. Berlin in his review of 67000 cases of foot tumors reported a malignancy rate of 1% and found that clinical feature of foot osteosarcoma are atypical [6].

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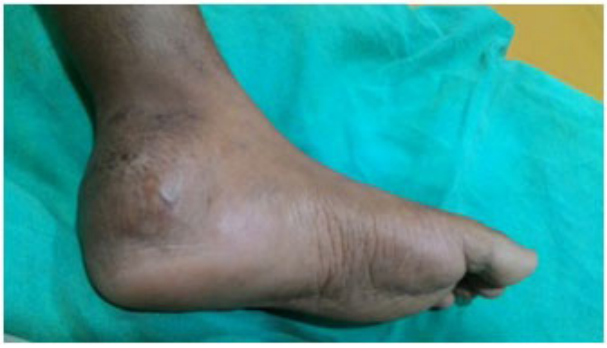

Figure 1. Showing swollen heel with dilated veins.

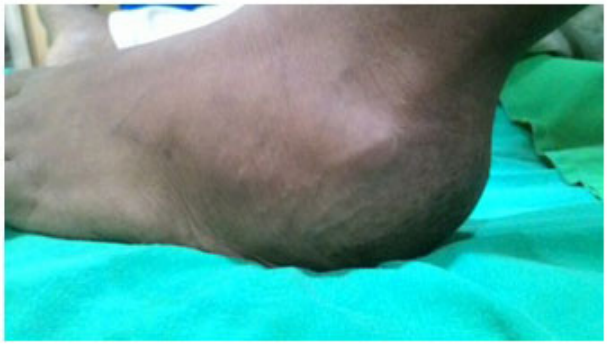

Figure 2. Showing swollen heel with dilated veins.

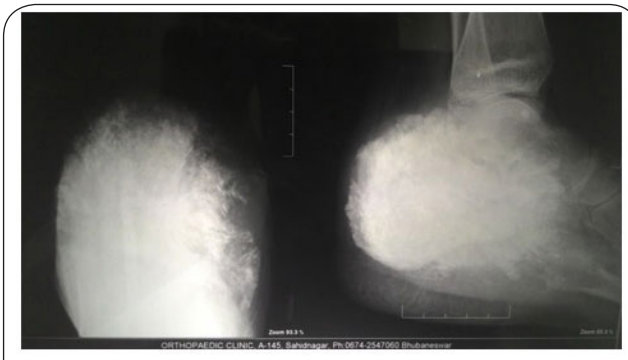

Figure 3. X-ray showing a dense sclerotic lesion involving the left calcaneum, with abundant periosteal reaction creating a sunburst appearance.

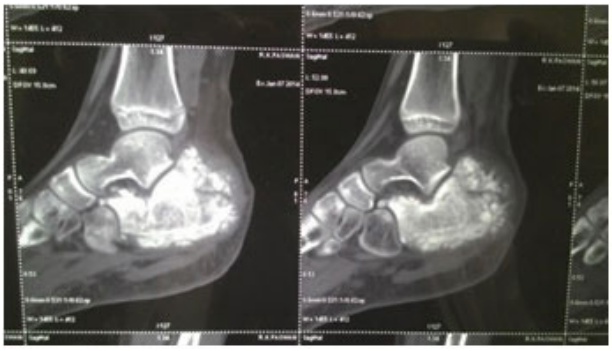

Figure 4. NCCT with 3D showing the diffuse periosteal reaction of calcaneum with abnormal soft tissue.

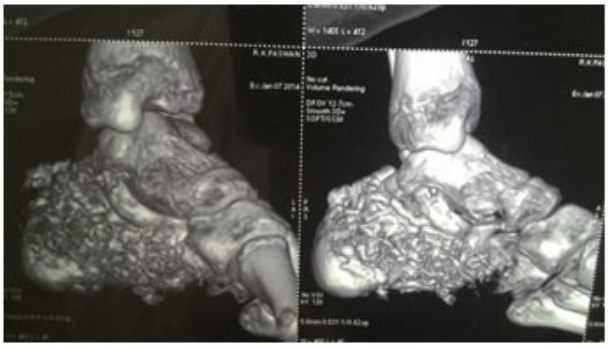

Figure 5. NCCT with 3D showing the diffuse periosteal reaction of calcaneum with abnormal soft tissue.

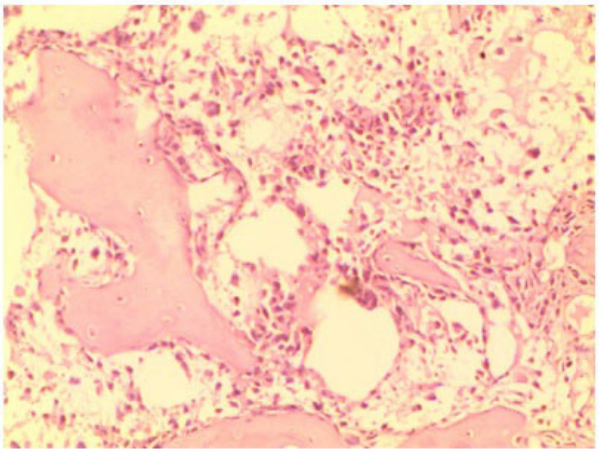

Figure 6. Histopathology section showing a malignant tumor infiltrating the bone with exuberant osteoid fornation. Individual cell shows moderate cytoplasm, enlarged irregular hyper chromatic to vesicular nuclei with coarse chromatin with prominent nucleoli.

Few literatures described osteosarcoma of foot as a distinct entity differing from their conventional counterpart having an incidence in older age groups and of lower grade in histopathology [2,3]. Most of reported cases in literature are in adults our case is rare being found in a adolescent male. As like of other types of bone tumors of foot, calcaneum is most common site for osteosarcoma of foot. In the series by Biscagalia et al., 6 out of 12 and in series by Choong et al., 11 out of 14 cases the primary site was calcaneum [2,3]. Syndromic osteosarcoma is found to be associated with Werner's syndrome, hereditary retinoblastoma, Paget's disease, Li-Fraumeni syndrome and Rothmund Thompson syndrome [9-12]. Being foot a tight compartment symptoms such as pain and swelling appears very early but rarity of occurrence and

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improvement of symptoms on conservative treatment lead to delayed diagnosis. In their review of 52 cases Choong et al., found that the average delay between presenting symptom and diagnosis varies from 1 to 24 months [3]. In our case also patient presented to us with a symptom since one year and two inconclusive FNAC reports. The differential diagnosis that should be considered are simple bone cyst, aneurysmal bone cyst, giant cell tumor, osteoid osteoma, osteoblastoma, and non-ossifying fibroma. The order of occurrence of malignant tumor in calcaneum as a primary site is osteosarcoma followed by Ewings sarcoma and chondrosarcoma [13,14]. Hence Biopsy is a must to distinguish calcaneal osteosarcoma from other two entities.

Like osteosarcoma of the long bones osteosarcoma of calcaneum depending upon the type of matrix production may be osteoblastic(commonest) , followed by fibroblastic and less common chondroblastic [3]. Some other reported subtypes of calcaneal osteosarcoma include low grade (intramedullary) type, telengiectactic type, periosteal type and small cell type. Osteoblastic type is the most common with dense sclerosis, ill defined margin and extra-compartmental bone production. Chondroblastic type differs from conventional type only in histology having chondroid cells and matrix [15]. Low grade intra-medullary type present as osteolytic lesion in x-ray with a sclerotic border and cortical affection having a relatively better prognosis. Telengietactic variety appears as osteolytic lesion with cystic fluid filled space like ABC. As there is no fascial barrier in the foot osteosarcoma of calcaneum easily get spread to adjacent compartment making management more difficult. CT and MRI are always needed to know involvement of compartments and for proper planning of management. Biscagalia et al., in their series of foot sarcoma found one half of their cases to be of low grade at presentation in contrast to Choong et al., who reported that most cases in their series at presentation were of high grade [2,3]. But all are in the same opinion that because of rarity of location and confusing symptoms, foot sarcomas present as referral case and at an advanced stage.

As it present at an advanced stages and poor compartmentisation of foot, limb salvage surgery has a restricted role in calcaneal osteosarcoma. There are sporadic case report of limb salvage surgery in calcaneal sarcoma mainly in low grade intramedullary type that respond well to neoadjuvant chemotherapy [16]. Response to chemotherapy of foot sarcoma is similar that of osteosarcoma of long bones [13,16]. Choong et al., has done below knee amputation in all their cases and reported that it is the satisfactory form of treatment with no evidence of local recurrence at 2 years follow-up [3]. In our case patient has undergone below knee amputation with no recurrence at 1yr follow-up and mobilized satisfactorily with below knee prosthesis.

Conclusion

Rarity of occurrence and atypical presentation of calcaneal osteosarcoma presents a challenge to orthopedics surgeon. Malignancy should be in mind while listing the differential diagnosis for a patient with chronic heel pain. Open biopsy is the standard investigation in scenario of doubt. As limb salvage for calcaneum is still in its infancy and tumor free margin is difficult to achieve because of poor compartmentisation and lack of fascial barrier, below knee amputation still the widely accepted treatment for long term survival of patient.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

Authors' contributions	BPS	CN	SP1	TKS	AKJ	SP2
Research concept and design	✓	✓	--	--	--	--
Collection and/or assembly of data	--	--	✓	✓	--	--
Data analysis and interpretation	--	--	--	--	✓	✓
Writing the article	✓	✓	--	--	--	--
Critical revision of the article	--	--	--	✓	✓	--
Final approval of article	✓	✓	--	--	--	--
Statistical analysis	--	--	--	--	--	✓

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