Osteosarcoma Of Calcaneum A Case Series Of Rare Tumour Presenting A Diagnostic Dilemma

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Article ID: WMC001273
Article Type: Case Report
Submitted on: 05-Dec-2010, 02:42:53 PM GMT Published on: 07-Dec-2010, 09:35:20 PM GMT
Article URL: http://www.webmedcentral.com/article_view/1273
Subject Categories: ORTHOPAEDICS
Keywords: Osteosarcoma, Calcaneum

How to cite the article: Chaudhary S, Singh D, Sen R, Deviraju C, Bhagwat K. Osteosarcoma Of Calcaneum A Case Series Of Rare Tumour Presenting A Diagnostic Dilemma. WebmedCentral ORTHOPAEDICS 2010;1(12):WMC001273

Source(s) of Funding: None

Competing Interests: None
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Abstract

Osteosarcomas of the calcaneum are rare. Owing to the rarity at this site, the diagnosis is often delayed or missed leading to the delay in treatment. We present a series of three cases of osteosarcoma of calcaneum, the difficulties in their diagnosis, their clinical, radiological and histological features that aid in the diagnosis and the management of these tumours.

Introduction

Osteosarcoma is the second most common primary malignant bone tumour that accounts for about 20% of all bone sarcomas and it commonly affects the appendicular skeleton[1]. The osteosarcoma of the foot is rare that constitutes about 0.8% of all osteosarcomas and calcaneum seems to be the favored site in the foot. Diagnosis is often missed or delayed owing to the rarity of the osteosarcoma at this location[2]. With early diagnosis and management, patients can expect a long term survival. Hence a great suspicion is needed while evaluating a mass arising from the calcaneum. Here we are presenting a series of three cases of osteosarcoma of calcaneum that presented a diagnostic confusion leading to the delay in treatment.

Case Report(s)

Case No. 1:-
A 17 years old male presented with painful swelling over lateral aspect of right ankle for last 9 months. Initially the swelling was small and increased progressively over last 9 months. There was no history of trauma, fever, any other swelling over the body. Patient was treated conservatively with analgesics and anti inflammatory drugs at a local hospital. Swelling was aspirated thought to be an inflammatory swelling but the diagnosis was not made.

On examination, 10x8 cm oval swelling was present over lateral malleolus. Swelling was firm in consistency with regular and well defined borders (Fig. 1). Local temperature was raised. There was restricted and painful dorsiflexion, eversion and inversion but normal plantar flexion.

Plain radiograph of the foot showed sclerotic lesions involving the entire calcaneum (Fig. 2).

MRI showed mildly expanded calcaneum with altered marrow signal associated with cortical breach at places and associated large soft tissue component on planter aspect of foot. Lesion was hypointense on T1W1 and hyper intense on T2W images (Fig. 3).

Hematological and biochemical investigations revealed no abnormality except that alkaline phosphatase was slightly raised (262.7 IU/L) Histopathological examination showed round to spindle shaped cells with hyperchromatic nuclei and moderate pleomorphism in the background of osteoid like matrix suggestive of osteosarcoma (Fig 4).

Bone scan and CT scan of chest and abdomen ruled out any distant metastasis.

In this case also the wide resection was not possible because of the involvement of entire heel flap. Below knee amputation was done. Post operatively chemotherapy was given.

There was no evidence of metastasis at follow up of one year.

Case No. 2:-
A 20-year old young male presented with pain and swelling in right foot for two months. He was treated by a general orthopaedic surgeon for 3 weeks as a case of plantar fascitis. Pain in the heel was the initial symptom; and 4 weeks later he also noticed a swelling in the posterolateral aspect of the foot (Fig 5). There was no history of trauma, fever or discharging sinus.

On Examination a diffuse, bony hard, non-tender swelling of around 6 x 4 cm was present on the poster lateral aspect of the ankle. The skin over the swelling was normal. The range of movements at ankle and sub-talar joints was restricted and painful.

Radiograph of the foot revealed a sclerotic lesion involving almost the entire calcaneum and new bone formation in soft tissues along the posterolateral aspect of the foot (Fig 6).

Hematological and biochemical investigations revealed no abnormality. Alkaline phosphatase, lactate
dehydrogenase, serum calcium, renal function tests and liver function tests were within the normal range. Open biopsy of the lesion was performed after 2 weeks of the presentation to our Institute. Specimen showed multiple fragments of white pearly tissue consisting of bone and soft tissue, which were firm in consistency. Microscopic examination showed sheets and whorls of tumor cells in a background of abundant osteoid and myxoid stroma. Tumor cells were small rounded with hyperchromatic nuclei and high mitotic rate (Fig 7). Based on these findings, the diagnosis of Osteosarcoma was confirmed. Three phase bone scan revealed no evidence of bony metastasis. Contrast enhanced tomographies of chest and abdomen ruled out any distant metastasis.

Wide margin resection was not possible because of the closed anatomic compartments of the foot. Tumor free heel flap could not be resected because of the associated posterolateral soft tissue component of the tumor, so a below knee amputation was planned in consent with the patient and his family. Below knee amputation was done within 2 weeks of its initial presentation. Patient received a full course of post operative chemotherapy. Simultaneously, patient was put on a program of aggressive physiotherapy to train the amputated stump for a period of 4 weeks. Patient was back to his work using a below knee prosthesis at the end of 4 months of his initial presentation to the Institute. At a follow up of 15 months, there was no evidence of any distant metastasis.

Case No. 3:-
A 30 years old female presented with complaint of heel pain on right side for six months. Pain was moderate and patient was unable to walk. Patient was diagnosed and treated as a case of planter fasciitis. Anti inflammatory medications and local steroid injection was given. Pt was not improved with 6 months of treatment. She developed swelling over heel and lateral aspect of foot. There was no history of trauma. On examination, swelling was present over heel and lateral aspect of foot which was firm in consistency (Fig.8). Radiograph of foot showed large calcified swelling at calcaneal spur. MRI showed swelling in muscles and soft tissue around the calcaneum on plantar aspect and hyperintense image on T2 weighted and STIR images, suggesting ?calcaneal osteomyelitis with involvement of surrounding nuscles and soft tissue or ? post traumatic marrow edema. Biochemical investigations showed no abnormality. Biopsy was taken from lateral aspect of foot and histopathological examination showed lobules of cartilaginous tissue at the periphery of which there was new bone formation by the tumour cells (Fig. 9). Patient was treated with chemo radiation of three cycles. Swelling decreased in size. Here also we were not able to do wide resection. Below knee amputation was done. Patient did fairly well with below knee prosthesis. Bone scan done at 11/2 years showed no recurrence or distant metastasis.

Discussion
Osteosarcoma is a high grade primary neoplasm that commonly occurs in the second and third decade of life and is common in the metaphyseal region of the bone. Commonest site is around the knee and accounts for approximately 50% of all cases[3, 4]. Osteosarcoma of foot is a rare entity. Some authors have suggested that osteosarcoma of foot may represent a distinct subgroup that differentiate it from conventional osteosarcoma as it is a low grade tumor and affects the older age group in contrast to its counterparts at the other sites[2]. Osteosarcoma of calcaneum have been associated with Hereditary Retinoblastoma, Werner syndrome, Rothmund Thompson syndrome and Li Fraumeni syndrome[3, 5, 6]. Pain and/ or swelling may be the initial presenting symptoms. Although these tumors tend to produce symptoms early because of the confined structure of the foot, their rarity may lead to delay in diagnosis. Further, the patients are frequently misdiagnosed with more common musculoskeletal problems during the initial visits. Pain may initially improve with conservative measures, giving a false sense of well being both to the patient and the physician. Osteosarcomas are commonly sclerotic in 45% of cases, purely lytic in 30% and mixed in remaining 25%[7]. There may be intense periosteal new bone formation and lifting of the cortex with formation of Codman’s triangle. Most important differential diagnosis for osteosarcoma of foot is Ewings sarcoma[7, 8]. Ewings sarcoma also presents in the same age group with usually similar radiographic features in the foot. So confirmation of the diagnosis can only be made with the help of open biopsy. Radiographic features in the form of large osteolytic / osteosclerotic changes with subperiosteal new bone formation should alarm the orthopaedic surgeon about the possible aggressive or malignant nature of the growth. Diagnosis can be further substantiated with
the MRI of the foot. In addition, MRI helps in the accurate estimation of tumor boundaries in relation to the surrounding structures. Although open biopsy has a greater risk of tumor spillage, hematoma and infection, it has a greater diagnostic accuracy[9]. In addition, possible disadvantages of core needle biopsy including a nondiagnostic biopsy, an indeterminate biopsy or a potential error in histological grade could be avoided[9]. Hence the open biopsy is the gold standard last step in the evaluation of the calcaneal mass which will affirm the histopathological nature of the growth. Delay in the diagnosis could be reduced by strictly following the above said protocol of investigations. Contrast Enhanced CT chest and abdomen are useful in further evaluating the osteosarcoma for its distant metastasis. Enneking had given a special consideration in classifying the malignant tumors of foot and treated the hindfoot and midfoot as a single compartment[10]. Osteosarcoma of the foot usually presents with the involvement of the surrounding compartments owing to the closed anatomic confines of the foot, lack of fascial barriers and thinner cortices. Thus it is almost impossible to achieve the radical oncological margins without performing a below knee amputation[11]. Even after extensive research of the literature we could find only three studies regarding foot salvage procedures for early stages of calcaneal osteosarcoma[12-14]. Thus, calcaneal resections and foot salvage surgeries are still in experimental stage in the current scenario; which further reinforces below knee amputation as the most preferred method of management of osteosarcoma of calcaneum[7, 8, 11]. The prognosis of these tumors still remains poor, with the overall survival rate depending on the specific type of osteosarcoma. The two year survival rate varies between 15- 20% for all osteosarcomas. However, with the advent of adjuvant chemotherapy, survival rates have increased to 60-70%[14].

Conclusion

In conclusion, the treating orthopaedic surgeon should keep the differential diagnosis of a malignancy while evaluating an osteolytic or osteosclerotic lesion of the calcaneum; which should be investigated appropriately. Open biopsy is the gold standard investigation in reaching at an early diagnosis and initiation of the appropriate treatment; which ultimately results in the long term survival of the patient.

References

Illustrations

Illustration 1

Figure 1 Clinical photograph showing swelling over lateral malleolus

Illustration 2

Figure 2 Plain radiograph of the foot showing sclerotic lesions involving the entire calcaneum.
Illustration 3

Figure 3 MRI showing mildly expanded calcaneum with altered marrow signal associated with cortical breach at places and associated large soft tissue component on planter aspect of foot.

Illustration 4

Figure 4 Photomicrograph showing round to spindle shaped cells with hyperchromatic nuclei and moderate pleomorphism in the background of osteoid like matrix suggestive of osteosarcoma.
Illustration 5

Figure 5. Clinical photograph of the right foot showing swelling on the posterolateral aspect of the calcaneum.

Illustration 6

Figure 6. Lateral radiograph of the right foot showing dense sclerosis of the entire calcaneum.
Illustration 7

Figure 7. Photomicrograph (X225) showing osteoid formation directly by the tumor cells and a focus of calcification at the upper left hand corner.

Illustration 8

Figure 8 Clinical photograph showing swelling over right heel
Illustration 9

Figure 9 Photomicrograph showing nodular lesions with aggregates and clusters of osteoblasts showing significant nuclear atypia and hyperchromasia at their periphery. (H&E x2)
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