

Osteosarcoma of The Talus: A Rare Location of the Tumour with Literature Review

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ABSTRACT

Osteosarcoma is the commonest primary malignant bone tumor. Most of them arises de novo in the metaphyseal area of the long bones. Osteosarcoma arising distal to the wrist or ankle is extremely rare and most often secondary to a premalignant process such as Paget disease. We reported a rare case of conventional osteosarcoma of the talus. A 40-year-old lady presented with painless left foot swelling for the past two years with gradually increasing in size within 6 months. Tru-cut tissue biopsy was reported as giant cell tumour. However histopathology of resected talus bone revealed that tumour was conventional osteosarcoma with focal features of giant cell rich osteosarcoma.

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Introduction

Osteosarcoma (OS) is the commonest primary malignant bone tumor. Most of them arises *de novo* in the metaphyseal area of the long bones, particularly the lower end of the femur, the upper end of the tibia, and the upper end of the humerus^[1]. OS arising distal to the wrist or ankle is extremely rare and most often secondary to a premalignant process such as Paget disease, radiation exposure and preexisting benign bone lesions^[2]. It usually occurs in patients between 10 and 25 years of age and is exceptionally rare in preschool children. Another peak age incidence occurs after 40, in association with other disorders. There is a slight male predominance. The large majority of OS arise within the medullary cavity, from which they extend into the cortex^[1]. Although there are some variants of OS, conventional type is the commonest.

When OS occurs in the foot bones, tarsal bones are commonest and calcaneus is the most involving^[3]. We reported a rare case of conventional OS in the talus with no history of previous preexisting lesion. A 40-year-old lady presented with painless left foot swelling for the past two years which gradually increasing in size within 6 months. Magnetic resonance imaging (MRI) report was suggestive of giant cell tumour (GCT) and Tru-cut tissue biopsy was also reported as GCT. However histopathology of resected talus bone revealed that tumour was conventional OS with foci of giant cell-rich OS area.

Case report

40-year-old, Malay lady presented to orthopaedic clinic for painless left foot swelling for the past two years. It was gradually increasing in size within 6 months. She had no history of trauma and any problem at left foot before the appearance of the swelling. On examination, there was a swelling over the medial and lateral aspect of left ankle, irregular in shape, firm to hard in consistency, slightly tender on palpation.

Plain radiograph of ankle (Figure: 1a) showed osteoblastic lesion occupying the talus. MRI (Figure: 1b) and tru-cut tissue biopsy were reported as suggestive of GCT; as the lesion was mainly composed of osteoclast-like multinucleated giant cells. Talus bone was excised together with tumour, navicular bone, lower end of tibia, upper part of calcaneus, surrounding soft tissue and overlying skin medially.

Grossly, specimen consisted of talus with tumor, navicular bone, lower end of tibia, upper part of calcaneus, surrounding soft tissue and overlying skin medially. On cut section (Figure 2), there was an infiltrating tumor occupying most of the talus and navicular bone invading anteriorly and posteriorly into soft tissue. Both superior and inferior surgical cut margin were involved by the tumor. Calcaneus bone and its articular cartilage were invaded by the tumor. Tibia and its articular cartilage were not invaded grossly. Lateral surgical margin was also involved by tumor grossly; however skin medially was not involved. Cut surface of the tumor was grayish white and firm in consistency. Small areas of hemorrhage were seen.

Histopathologically, there was an infiltrating bone forming tumor originating in the talus. Tumor was mainly composed of spindle to epithelioid looking pleomorphic cells. Mitoses were frequent varying 5-20/10HPF depending on cellularity. Most of the area in between the tumor cells showed pink, amorphous lace like pattern of osteoid tissue. (Figure: 3a) Osteoclast like multinucleated giant cells were seen admixed with tumor cells. (Figure: 3b) Areas of dense collagenization were also seen in the stroma. Small cystic-like spaces with haemorrhages were also seen. With those histopathological findings, we concluded that it was conventional OS, originated in the talus bone.

After excision of the tumour, CT (computerized tomography) thorax showed three lung nodules suggestive of metastasis. Patient was on chemotherapy and radiotherapy; responding well with rehabilitation.



Fig. 1: (1a. Plain radiograph) & (1b. MRI) showed heterogeneous lesion (asterisk) involving the whole talus with surrounding soft tissue mass.

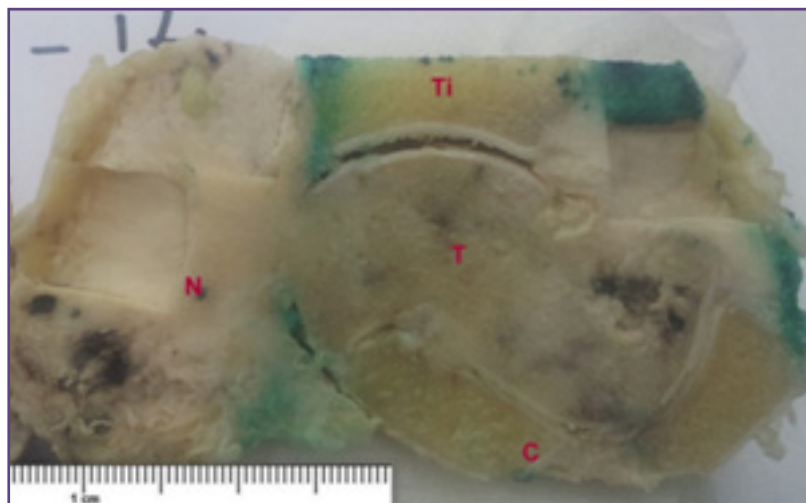


Fig. 2: Tumour arising in talus (T) invading navicular bone (N) and calcaneus (C). Tibia (Ti) and its articular cartilage are not invaded. Cut surface of the tumour is solid, grayish white and firm to hard in consistency.

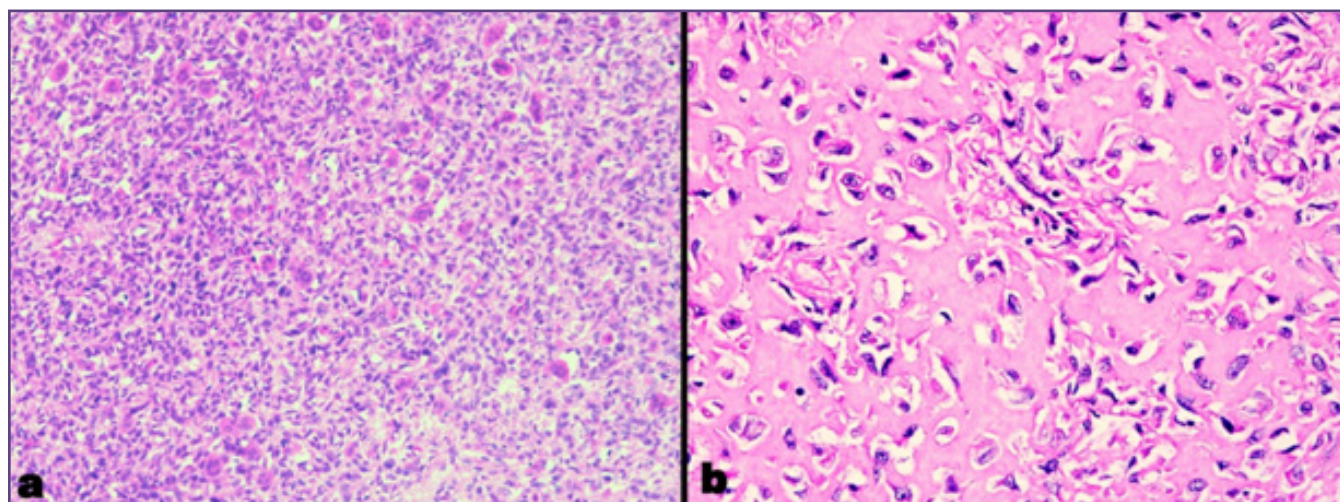


Fig. 3: (a). Giant cell rich area of the tumour which mimics GCT. (H&E 200X) (b). Pleomorphic tumour cells and pink amorphous tumour osteoid forming a lace-like pattern. (H&E 400X).

Discussion

OS arising distal to the wrist or ankle is extremely rare and most often secondary to a pre-malignant process such as Paget disease [2]. In a study of 52 cases of osteosarcoma of the foot, tarsal bones are commonly involved. Among them, calcaneus is the commonest followed by the talus and navicular bone [3]. In a study of OS of hands and feet, 62 % of the OS presented in the metacarpals and 23 % in the phalanges, and only two cases occurred in the carpal bones. Distribution in the foot was 56 % in tarsal bones, 33 % in metatarsal bones and 11 % in phalanges [4]. In a study of 1929 cases of OS, only 12 cases were located in the foot with the commonest in the calcaneus followed by the talus [5]. Among the tumours of the foot and ankle, GCT is the commonest [6].

Most of OS affects children and adolescents. However, some of the case studies on OS affecting the foot showed that most of the patients were older than 30 years [5,6]. There were some reported cases of talar OS in elder age without any underlying pathology [7]. In addition, patients with OS of the foot commonly have a long delay from the time of onset of symptoms to diagnosis, with the mean interval greater than 2 years [5]. Patient in this case was middle age woman and onset was slow without any underlying pathology.

In a study of OS of hands and feet, 3 cases were seen in the talus out of 27 cases of osteosarcoma feet. That study also found out that patients with OS of the distal extremities are older, have a different gender distribution, and differ

in symptoms, history and grade of malignancy [4]. Most of the OS of hands and feet present with pain with or without swelling and the average duration of symptoms were 13 months [4]. Some of the cases of OS foot may present with painful swelling which lead to misdiagnosis of inflammatory condition [8]. Clinical findings of most of OS of the talus are atypical and it leads to misdiagnosis and delayed correct diagnosis [5]. This case presented with painless swelling for 2 years duration.

In this case, Tru- cut biopsy before excision of the tumour was suggestive of GCT as it was mainly composed of multinucleated giant cells with few mononuclear cells. The biopsy was seemed to be taken from the site of giant cell-rich area of the tumour. When we correlated with plain radiograph images, it was osteoblastic lesion rather than osteolytic lesion. The case was not associated with any pre-malignant bone lesion or prior irradiation.

Generally, histopathological diagnosis of OS is mainly based on identification of osteoid and/or bone (calcified osteoid) produced directly by the tumor cells without interposition of cartilage [1]. Immunohistochemical expression of specific proteins produced by tumour cells of OS is supportive in the diagnosis. Extracellular matrix metalloproteinase inducer (CD147) is expressed in human osteosarcoma tumor tissues [9]. Proteins specifically associated with bone metabolism such as osteonectin, osteocalcin, osteopontin and bone morphogenetic protein have been identified immunohistochemically in the cells of osteosarcoma and may be useful in the differential diagnosis of OS [1]. For differentiation between OS and GCT, specific proteins of GCT are useful. A study on GCT showed that strong immunohistochemical expressions of osteoprotegerin and osteoprotegerin ligand are seen in the giant cells and background stromal cells [10]. However, these special immunohistochemical stains are not routinely used and not performed in this case. GCT is distinguishable from giant cell-rich OS in haematoxylin and eosin stained sections. GCT does not produce typical lacy tumour osteoid and their stromal cells do not exhibit pleomorphism.

In conclusion, OS of the talus bone is rare. Accurate diagnosis can be complicated by subtle clinical features. Thorough radiological correlation in histopathological diagnosis is very important. Representative tissue biopsy is essential to provide thorough histopathological examination to reach correct diagnosis.

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Competing Interest

None

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