CASE REPORT

Osteochondromyxoma of Talus

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Osteochondromyxoma is a rare bone tumour. Bone tumours of the talus are also uncommon, and accounts to be between 8% to 23% in tumours of the foot. A 28-year-old man presented with chronic right ankle pain. He had underlying left knee ligament and meniscal injury. Special examination tests for ligament injury were negative. Magnetic Resonance Imaging (MRI) revealed a benign bone lesion of talus with reactive oedema of sinus tarsi. Excision of lesion was done and subsequent histopathological examination confirmed the diagnosis of osteochondromyxoma.

INTRODUCTION

Osteochondromyxoma is a benign chondrogenic bone tumour that is extremely rare1, 2. Meanwhile, bone tumours of the talus are also uncommon and accounts to be between 8% to 23% in tumours of the foot3. The common benign bone tumours of the talus are osteoid osteoma and intraosseous ganglion cyst1. Chondroblastoma, fibrous dysplasia, primary aneurysmal bone cyst, giant cell tumour, chondromyxoid fibroma, osteochondroma, osteoblastoma, intraosseous lipoma and epithelioid haemangioma are other less common differentials for benign growths of the talus4. To the extent of our knowledge, no case report was found on osteochondromyxoma of the talus. Herein, we present a case of osteochondromyxoma in talus of a young male patient.
CASE PRESENTATION

A 28-year-old man was under orthopaedic clinic follow up for left anterior cruciate ligament complete tear with lateral meniscus injury. He had history of sports-related injury of the left knee in 2015. During one of the reviews, patient complained of right ankle pain for 7 months and was unable to bear weight for long duration. The pain score was 7/10. It was also associated with instability. However, no recent history of fall or trauma claimed by the patient. On examination of the right ankle, tenderness was noted at the lateral aspect. There was no associated swelling of the joint. Range of movement was full, however pain noted on inversion and eversion of the ankle. The anterior drawer test and talar tilt test were negative. He was also noted to have worsening ankle pain post-operatively after undergoing left anterior cruciate ligament reconstruction. It required him to use ankle support cast for walking.

Plain radiograph of the ankle was performed showing small lytic lesion with sclerotic border at lateral process of talus, suspicious of osteochondral defect of talus (Figure 1). Therefore, he underwent MRI for further assessment of the lesion.

Figure 1 AP and lateral view of right ankle showing lytic lesion with sclerotic border at lateral process of talus (white arrow)

MRI of right ankle revealed a well-defined intraosseous lesion at lateral process of talus with extension to the superior part of sustentaculum tali. It returned hypointense signal in T1-weighted images, intermediate signal intensity in T2-weighted images and proton density images, hyperintense signal intensity in proton density fat suppression images and peripheral rim enhancement in post Gadolinum sequences. There were also adjacent bone oedema and heterogenous signal intensity of adjacent sinus tarsi suggestive of reactive oedema. The ankle ligaments return normal signal intensity. The findings were suggestive of benign bone lesion of talus.
Figure 2 T1 weighted MRI showing coronal view (a) and axial view (b) of right ankle with hypointense lesion at the lateral process of talus (white arrow).

Figure 3 T1 Weighted MRI showing post gadolinium coronal view (a) and axial view (b) of right ankle with peripheral rim enhancement of talar lesion (white arrow).
The lesion was then excised and sent for biopsy. Histopathological examination showed fragmented tumour tissue composed of generally patternless sheets of spindle cells separated by basophilic myxoid stromal. The cells are arranged in lobules in some areas. The cells were polygonal, elongated and stellate shaped with hyperchromatic small nuclei. The histological findings were compatible with osteochondromyxoma of bone (Figure 7).

**Figure 4** T2 Weighted MRI showing axial view of right ankle with intermediate signal intensity lesion at talus (white arrow)

**Figure 5** Proton density weighted MRI showing coronal view of right ankle with intermediate signal intensity lesion at talus (white arrow)

**Figure 6** Proton density fat suppression (PDFS) weighted MRI showing coronal view (a) and axial view (b) of right ankle with hyperintense signal intensity lesion at talus (white arrow)
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**HISTOPATHOLOGY REPORT:**

**MACROSCOPY**
Specimen labelled as Tissue.
Multiple pieces of tan tissue measuring 30mm in aggregate diameter. Entirely submitted in 1 block.

**MICROSCOPY**
Section shows fragmented tumour tissue composed of generally patternless sheets of spindle cells separated by basophilic myxoid stromal. In areas, the cells are arranged in lobules. The cells are polygonal, elongated and stellate shape with hyperchromatic small nuclei. No malignancy.

**INTERPRETATION**
Osteochondromyxoma.

Post-operatively the patient condition has improved. During the subsequent one-month review there was no further complaint of pain and range of movement was preserved.

**DISCUSSION**

Bone tumours of the foot represent only 3% of osseous tumours. Bone tumours of talus represent 8% to 23% of the tumours of foot. The rare bone tumours of the talus that have been reported include giant cell tumors, aneurysmal bone cyst, intraosseous lipoma, chondroblastoma, osteoblastoma and osteochondroma. On the other hand, osteochondromyxoma is an extremely rare tumour and only a few cases have been reported before. It is a benign chondrogenic bone tumour that demonstrates both osteoid and chondroid production. It arises in approximately 1% of patients with Carney complex and has also been associated with lentigines and other unusual disorders as well. The usual site of involvement includes tibia, radius and sinonasal bones. There are also other non-English literatures that have reported osteochondromyxoma of rib, chest wall and spine. None was reported arising from the talus as in our case. It originates from cortices and is known to be a locally aggressive tumour despite of the benign nature, causing destructive growth with extension into soft tissue. It presents as a painless mass with additional symptoms due to oedema and mass effect. It is associated with discomfort symptoms depending on its site and size. In correlation with our case, the pain attributed by reactive changes and irritation of sinus tarsi.

On radiographic imaging, it is usually a well circumscribed mass which may also show destructive or mineralized signs. It can also show expansion of the affected bone area with mixture of lucent and sclerotic regions. MRI usually reveals increased signal in T2-weighted imaging. Differential diagnosis includes chondromyxoid fibroma and chondrosarcoma with myxoid changes. Histologically, it is characterized by cells containing eosinophilic cytoplasm and nuclei arranged in rows. It consists of sheets with lobular areas, polymorphic cells, chondroid, osteoid and hyaline bands. Light microscopy usually shows mixture of mesenchymal cells, basophilic...
myxoid material and mucopolysaccharide ground substance. There is also presence of osteoid and bone, immature and mature cartilage, hyaline fibrous bands and nodules as well as collagen fibres scattered within the tumour. The cells are usually organized in sheets, either well-defined or disorganized microlobular or macrolobular patterns. Most cells found in microscopy are polygonal, stellate and bipolar. The nuclei of the cells are well preserved, chromatic and vesicular with small nucleoli.

Osteochondromyxoma has a good prognosis with complete excision. However, local recurrence is common and usually occurs at sites where complete excision is difficult.

CONCLUSION

A case of osteochondromyxoma of talus is presented for the first time in our hospital. The tumour arises from talus causing non-specific clinical symptom, therefore, imaging plays a very important role for characterisation of lesion. Even though osteochondromyxoma of talus is an extremely rare diagnosis, it should be included in the differential diagnosis of bone tumour of talus.

CONFLICT OF INTEREST

The authors declare that they have no competing interests in publishing this case.

CONSENTS

Written informed consent was obtained from the patient to publish the case with its related pictures. A copy of the written consent is available for review by the Chief Editor.
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