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BENIGN FIBROUS HISTIOCYTOMA OF CALCANEUM - A RARE CASE REPORT

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ABSTRACT

Introduction: Benign fibrous histiocytoma of the calcaneum is an extremely rare neoplasm and it is essential to differentiate it from other common giant cell lesions of bone particularly giant cell tumour of bone.

Case Presentation: We present a case of a 38 year old female with an osteolytic lesion in the left calcaneum with a clinical and radiological diagnosis of giant cell tumour of the bone. The definitive diagnosis of benign fibrous histiocytoma was confirmed by histopathological examination and P63 immunomarker negativity.

Conclusion: This case is presented due to the rarity of its occurrence in the calcaneum and to stress the significance of using p63 as an immunomarker in differentiating from giant cell tumour.

Keywords: Benign fibrous histiocytoma, Giant cell tumour, P63.

INTRODUCTION

Benign fibrous histiocytoma (BFH) is a soft tissue tumour of fibrous histiocytic type rarely occurring in the bones, especially the calcaneum. It is a spindle cell neoplasm of the bone and needs to be differentiated from non-ossifying fibroma and giant cell tumour of bone. ^[11] Very few cases of benign fibrous histiocytoma arising in the calcaneum have been reported in the literature. We report a case of benign fibrous histiocytoma in the calcaneum occurring in a middle aged female and the role of P63 in differentiating it from the giant cell tumour. ^[2]

CASE REPORT

A 38 year old female presented with pain in the left foot mainly on weight bearing which gradually increased in intensity for the past six months. There was no significant swelling or other relevant history of injury noted. Clinical examination of the foot revealed bony tenderness with the X-ray showing an osteolytic lesion in the left calcaneum. Haematological and biochemical parameters were within normal limits. In view of the radiological appearance, a diagnosis of giant cell tumour of bone / chondromyxoid fibroma was considered. [Figure 1]

Under spinal anaesthesia, curettage of the osteolytic lesion was done and sent for histopathological examination. Histopathology revealed only tiny fragments of bony tissue with sheets of foamy histiocytes. There were no osteoclastic giant cells to suggest giant cell tumour or chondromyxoid fibroma. Since the material was very minimal, a repeat curettage was advised and planned for a confirmatory diagnosis.

Under spinal anaesthesia, repeat curettings of the entire lesion was done and sent for histopathological examination. The intraoperative and postoperative period was uneventful. Macroscopically, multiple yellowish and grey white bony and soft tissue fragments measuring 3x2x1.5 cm in aggregate were seen. [Figure 2]

Microscopic examination revealed a lesion composed predominantly of foamy histiocytes and scattered spindle cells arranged in a storiform pattern. [Figure 3] There were osteoclastic giant cells scattered throughout but not seen uniformly in a spatial relationship as in a giant cell tumour of bone. A diagnosis of benign fibrous histiocytoma was made and confirmed by negative immunostaining for p63. [Figure 4] Patient came for follow up and is doing well.

DISCUSSION

Benign fibrous histiocytoma is a spindle cell neoplasm with varying degrees of spindle cells, foam cells and multinucleated giant cells. They occur very rarely within bones and approximately 86 cases have been reported in literature with this case being only the second case to be reported in the calcaneum, a very unusual site. ^[1]

Benign fibrous histiocytoma, a tumour known to occur in the dermis and soft tissues, comprised about ten cases out of 11087 skeletal tumors studied by Dahlin and Unni. ^[3] The cases were adults between the ages of 23 to 60 years and the male: female ratio was 4:3. In a series reported by Grohs et al, a median age of 28 years and male: female ratio of 4:6 was noted.^[4]

Many authors actually suggest that it is a non ossifying fibroma, and the location of the lesion with clinical history of pain and the age group is essential in making a diagnosis of BFH.^[2]

Local pain is the main symptom in benign fibrous histiocytoma in contrast to nonossifying fibroma and the X-ray shows an osteolytic lesion with sharply defined sclerotic borders in a typical case of benign fibrous histiocytoma. In the present case reported there was no thick sclerotic rim.^[5]

Nonossifying fibroma is usually an incidental radiological finding and is seen in patients younger than 20 years. It is unusual after skeletal maturation. The most common site is the distal femur and proximal tibia. Benign fibrous histiocytoma occurs in adults and usually in the long bones and pelvis. In a case series comprising ten benign fibrous histiocytomas of bone, the common sites were femur, pelvis, humerus, tibia, fibula, ribs and spine.^[3]

Benign fibrous histiocytoma also needs to be differentiated from the common clinical and radiological diagnosis of giant cell tumour of bone as in our case. Most neoplasms of the bone may contain giant cells but uniform spatial arrangement of osteoclastic giant cells with mononuclear stromal cells is critical in making a diagnosis of giant cell tumour. Lesions with osteoclastic giant cells and spindle cells if present in flat bones also need be differentiated from to hyperparathyroidism and aneurysmal bone cyst which have other distinctive histological features. [5]

The diagnosis of giant cell tumour needs to be consistered strongly as collections of foam cells dominate the picture and mononuclear cells may become spindled and have even a storiform pattern, in which case a mistaken diagnosis of benign fibrous histiocytoma can be made.^[2] A lesion with all the clinical and radiological features of giant cell tumor has to be diagnosed as giant cell tumour even if the predominant pattern is of benign fibrous histiocytoma and only minor foci shows osteoclastic giant cells.^[2]

In such cases p63 immunostaining can be of immense help in differentiating benign fibrous histiocytoma (p63 negative) from giant cell tumour (p63 positive).^[6] p63 is a p53 homolog expressed in normal epithelial tissues and in epithelial malignancies. It is mainly used as a marker for mammary myoepithelial cells and prostatic basal cells and recent studies have indicated moderate nuclear staining in the stromal cells of giant cell tumor of bone. Non ossifying fibroma shows less than 10% staining for p63 and generally most of the soft tissue tumors do not express p63.^[7] The behaviour of benign fibrous histiocytoma is benign in contrast to the locally

aggressive behaviour of giant cell tumor of the bone. $\ensuremath{^{[8]}}$

CONCLUSION

Benign fibrous histiocytoma can occur in the calcaneum and the X-ray picture may be atypical as in our case and the significance of using p63 as an immunomarker in differentiating from the clinical and radiological diagnosis of giant cell tumour is stressed in doubtful cases where microscopic features are equivocal. This case is presented due to the rarity of its occurrence in the calcaneum, as to the best of our knowledge, this is the second case to be reported in the calcaneum in literature.

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PICTURES



Figure 1. X ray foot with ankle joint showing an osteolytic lesion with a thin rim of sclerosis in the calcaneum.



Figure 2. (a) Intraoperative picture of the lesion (b) Picture of the entire currettings material.



Figure 3. Microscopic view of the lesion showing storiform pattern of spindle cells with scattered osteoclastic giant cells. (a) H and E x100 (b) H and E x400



Figure 4. (a) Immunostaining for P63 showing negative staining in this case x400 (b) Control slide (Giant cell tumour) showing P63 positivity x400.