Giant Cell Tumor with secondary aneurysmal bone cyst of second Metacarpal: A case report and review of literature

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Abstract

Only about 2% of Giant Cell Tumors occur in the hand. Furthermore, metacarpal involvement is much less common than the phalangeal involvement. Giant cell tumor with secondary aneurysmal bone cyst component in metacarpal is a rare presentation. We report a case of Giant Cell Tumor (GCT) with secondary aneurysmal bone cyst of second metacarpal in eighteen years old female treated with enbloc resection and fibular strut grafting.

Keywords: Giant Cell Tumor with secondary aneurysmal bone cyst, fibular graft, metacarpal.

Introduction

Giant cell tumor or osteoclastoma is generally a benign, locally aggressive tumor with a high tendency for local recurrence and occurring mostly during the second and third decade of life [1]. Only about 2% of giant cell tumors occur in the hand and very rarely in metacarpal [2-4]. The recurrence rate being higher when these occur in hand wherein, it occurs in a relatively younger age group and shows a more aggressive behaviour [5] The occurrence of secondary aneurysmal bone cyst in a pre-existing giant cell tumor is about 14%, [6] furthermore; such an occurrence in a metacarpal is very rare.

Case Report

An 18 year old female student presented to us with chief complaint of a gradually increasing swelling with insidious onset over the second metacarpal of Left hand with persistent and progressive pain over a period of the last 6 months. On examination a 6X5 Sq. cms, oval shaped swelling was noted over the dorsum of the left hand all along the 2nd metacarpal. The Swelling was tender, non-mobile, firm in consistency, with well-defined borders and confined to second metacarpal. (Fig 1) The overlying skin was normal. There was no history of trauma or any other constitutional symptoms. Patient was then evaluated with appropriate radiographs and lab Investigations. The radiograph revealed an eccentric lytic lesion of the II metacarpal, multiseptate with soap bubble appearance suggestive of a Campanacci [7] grade 2 giant cell tumor. (Fig 2)

Surgical Procedure

With patient lying supine, under GA, and with left forearm and hand resting over an Arm board under Tourniquet control, about 10 cms of incision was applied over the dorsal aspect of the II metacarpal of left Hand. The dissection was done under loupes while taking care to raise thick flaps and taking care of extensor tendons and neurovascular bundles. The tumor was found to be well encapsulated without any involvement of the surrounding soft tissues. (Fig 3 a & b) Enbloc resection of entire second metacarpal with the encapsulated tumor was done (Fig 4 a). In gross appearance the tumor showed a greyish brown mass with isolated areas of necrosis with multiple loculi and uniformly thickened cyst wall. (Fig 4 b)

The bone gap was measured and appropriate sized Fibular strut graft was harvested. Multiple drill holes were made in the strut after establishing the medullary canal to ensure better incorporation of the graft. The defect was reconstructed with this fibular graft and fixed with K-wire to the proximal phalanx & carpal bones under image guidance and augmented with tricortical autologous iliac crest graft (Fig 5 a & b). The tissues were closed over the graft taking sutures through the drill holes. The wound was then closed in layers.

The tumor tissue was sent for histopathological examination which
revealed multilocular cystic lesion, with large vascular spaces with fibrous septae and numerous benign Osteoclastic giant cells. The impression was Giant cell tumor with secondary aneurysmal bone cyst component. (Fig 6 a & b)

**Post Operative care:**
The patient was given a below elbow POP volar slab with the hand in functional position. Suture removal done at the end of 2 weeks. Gentle intermittent passive mobilisation of the IP joints is continued and progressed gradually to active ROM over a period of 8 weeks. The K wire was removed at the end of 10 weeks and the POP slab discontinued. Active hand and wrist mobilization was started. The fibular graft was fully taken up at 3 months. PIP and DIP Joint movements of the Index finger were well maintained and patient was able to do all activities of her daily living. At 5 years of follow-up, there has been no sign of recurrence both radiologically (Fig 7) and clinically (Fig 8a-f) and patient is able to do all her activities of daily living along with her occupational work of tailoring.

**Discussion**
Cooper [8] first reported giant cell tumors in the 1818. In 1940, Jaffe et al began a systematic analysis of those lesions that contained giant cells, emphasizing the additional histological characteristics of the giant cell tumour thereby differentiating it with other giant cell containing neoplasms [9]. Giant cell tumor accounts for 5 % of all primary bone tumors, however; the rate is 20 % in southern India (Andhra Pradesh) and china [10]. 85-90 % of the cases of GCT arise denovo from the epiphysis of long bones and eventually involve the metaphysis predominantly in the age group of 20-40 years [11]. These tumors are much less common in skeletally immature patients and there is a definite female predominance with a ratio 1:1.5. Giant cell tumors are mostly solitary in nature, however 1-2 % are multicentric in nature and about 5 % are malignant [2]. Aneurysmal bone cyst (ABC) is a benign cystic lesion of bone that is composed of blood-filled spaces separated by connective tissue septa containing fibroblasts, osteoclast-type giant cells, and reactive woven bone [12]. ABC may be either primary or secondary. Primary ABCs are those which arise de novo. A secondary ABC develops in association with other neoplasms, most commonly GCT of bone, osteoblastoma, chondroblastoma, and fibrous dysplasia. GCT under-going a intra-lesional hemorrhage and resulting a resorptive osteolysis of surrounding bone. This could be the basis of the aneurysmal bone cyst formation and therefore, considered as secondary [13]. The typical appearance of GCT is best demonstrated in plain radiographs is

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*Figure 1*: Preoperative Photograph showing the swelling of 2nd metacarpal.

*Figure 2*: Photograph showing preoperative radiographs anteroposterior and lateral views.

*Figure 3*: (a & b): Photograph showing Intraoperative appearance of the tumour.

*Figure 4*: (a & b): Photographs showing enbloc excised tumour mass and multilocular appearance of the tumour.

*Figure 5*: Photograph showing preparation of the fibular strut and k wire fixation of the fibular strut in the defect.

*Figure 6*: Histopathological images of the tumor showing coexisting features of Giant cell tumour and Aneurysmal bone cyst.
diagnostic. The lesion is purely lytic, expansive, soap bubble in appearance and eccentrically located in epiphysis of long bones. Periosteal reaction is seen in case of pathological fracture [14].

CT-scans help to determine exact amount of cortical destruction, joint surface and determine the optimal location of the cortical window for the purpose of curettage. MRI may help in determining the extent of lesion mainly in the soft tissue or fluid levels if any [7].

Histologically GCT is composed of multinucleated giant cells, 40-60 nuclei per cell in a sea of mononuclear stromal cells. Areas of storiform spindle cell formation, reactive bone formation of foamy macrophages may be seen. The stroma of most GCTs is vascular and contains numerous thin-walled capillaries, often with small areas of haemorrhage [6] These lesions may be associated with secondary aneurysmal bone cyst (ABC) formation but also contain solid areas with the typical histologic appearance of GCT (Fig 6 a & b) . The identification of these solid areas of GCT allows differentiation from primary ABC, which contains only hemorrhagic cystic regions [6]. The various treatment modalities described in literature are simple curettage, curettage with bone grafting, en bloc resection with reconstruction of joint surface using silastic prosthetic implants, amputation, arthrodesis, radiotherapy, chemotherapy & embolization [10, 14, 15]. The metacarpalphalangeal joint reconstruction can be achieved by metatarsal substitution with a combined iliac crest graft, nonvascularised fibular graft or silastic prosthetic replacement. The local recurrence rate can be as high as 90 % in hand [15]. In our case since the whole metacarpal was involved without soft tissue involvement, we performed an en bloc resection followed by metacarpal reconstruction using fibular autograft with autologous iliac crest grafting. The patient showed complete remission at a 5 year follow-up and has gone back to her activities of daily living and professional activities.

References