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Bifocal metachronous giant-cell tumour of ulna and distal radius

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CASE REPORT

Bifocal Metachronous Giant-Cell Tumour of Ulna and Distal Radius

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ABSTRACT

A 36 years old gentleman had giant cell tumour (GCT) in distal ulna for which he underwent resection of distal half of ulna but developed recurrence of the tumour. Following proper evaluation (grading and staging), he underwent wide margin excision of tumour including removal of distal three-fourth of ulna and reconstruction by free vascularised osteocutaneous fibular transfer. The distal reconstructed fibula was stabilized with extensor carpi ulnaris (ECU). Two years after the surgery, he developed a metachronous GCT lesion in ipsilateral distal radius for which he had curettage and bone grafting with preservation of articular surface.

Key Words: *Giant cell tumour. Metachronous tumour. Recurrence. Reconstruction.*

INTRODUCTION

GCT accounts for approximately 20% of all primary bone tumours in Southeast Asian region.[1] It is found most often in the epiphyseal ends of long bones, especially the distal femur, proximal tibia, and distal radius. GCTs occurrence at the distal end of the ulna is extremely rare, accounting for 0.45 - 3.2% of all the cases of GCTs.[2] Giant cell tumours usually are solitary lesions; however, 1 to 2% may be synchronously or metachronously multicentric.

Though GCT of ulna is extremely uncommon,[3] the bifocal metachronous involvement of radius and ulna are not reported.

CASE REPORT

A 36 years old gentleman, manual labourer, developed swelling in his left distal forearm in January 2009, which was diagnosed as GCT of distal ulna. For that, he underwent resection of distal half of ulna outside of our institution. His histopathology report confirmed GCT. He presented in the authors' clinic for the first time in January 2010 with recurrent swelling in his left forearm. There was no history of any other swelling in the body, fever and weight loss.

Physical examination revealed a large swelling of 12 x 8 cm in the mid forearm with surgical scar mark on ulnar border. The swelling was firm with non-adherence skin to the underlying mass. The range of motion (ROM) at wrist and elbow was restricted and painful. Neuro-vascular status was intact and axillary lymph nodes were not palpable.

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Blood examination was within normal limits. Plain X-ray showed large soft tissue mass shadow in volar aspect of forearm. Only proximal one-third ulna was visible with eroded, ill-defined edges merging into soft tissue mass. MRI demonstrated a soft tissue well circumscribed mass of 12 x 7 x 5 cm in volar compartment of left forearm with pressure effect on flexor muscle sparing neurovascular structures. Distal end of the remaining ulna was eroded with signal changes. A clinical diagnosis of recurrent GCT was made and confirmed with biopsy. His work up for staging revealed isolated ulnar lesion with no metastasis.

Considering a locally aggressive stage III benign tumour, he underwent wide margin excision of the tumour, resection of diseased ulna and involved surrounding muscles. Anterior interosseous nerve was sacrificed due to involvement in the mass. Approximately 4 cm of proximal ulnar stump was retained saving elbow joint. Tissues from deep anterior compartment and marrow from the proximal ulnar osteotomy site was sent for frozen section which were found tumour-free. His ulnar defect was reconstructed with free vascularised osteo-cutaneous fibular graft which was harvested from ipsilateral leg and anastomosed with ulnar artery and veins. Fixation of the fibular graft with ulnar stump achieved with reconstruction plate and distal ulna was stabilized with sling of ECU (Figure 1).

At one year follow-up, his wrist was pain-free and had fairly good range of motion at wrist and elbow. X-rays showed union at graft-ulna site with no other abnormal findings. At 2 years follow-up, he developed new focus of lytic lesion in distal radius which was absent in previous imaging (Figure 2). CT scan wrist with contrast showed expansile lesion of 3.7 x 1.8 x 1.9 cm lesion in distal radius with breech in cortex at interosseous border and soft tissue extension but preservation of articular surface. Work-up for staging did not reveal metastasis.

He underwent curettage of the distal radius with high speed burr and instillation of alcohol and phenol. During

Figure 1: Pre-operative X-rays at the time of presentation before wide margin excision. Postoperative X-rays after excision of tumour and reconstruction with vascularised free fibula.

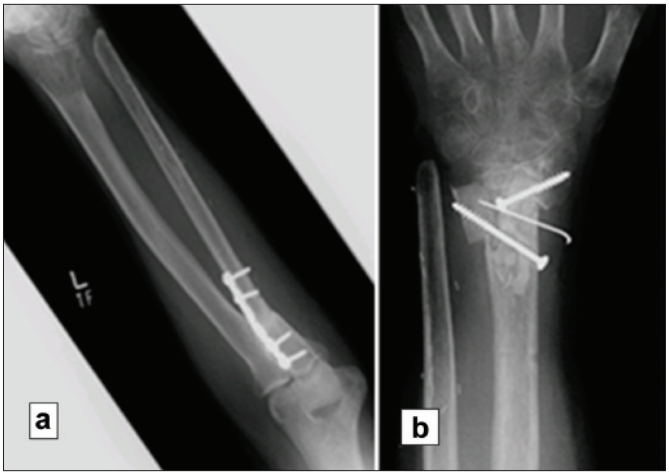

Figure 2: (a) New focus of tumour in distal radius at 2 years follow-up after reconstruction with osteocutaneous flap. **(b)** Curettage of distal radius GCT lesion with reconstruction by bone graft.

curettage, he developed fracture in the cortex, hence reconstruction was done with iliac crest cortico-cancellous graft and stabilization with screws, k-wires and AO external fixator. Fixator was removed after 6 weeks and gentle ROM started. Now around one year after surgery, he is pain-free with adequate ROM and no recurrence.

DISCUSSION

Multicentric giant-cell tumour may be either synchronous which is characterized by lesions that are remote from one another but are discovered within a short period of time and at similar stages of development or metachronous that are characterized by lesions that occur at different times and in different locations. Metachronous tumours may be metastatic or may represent a second independent focus of disease.[4] This case showed bifocal metachronous GCT.

Multicentric giant cell tumour is rare and accounts for < 1% of all cases of giant cell tumour of bone.[5] The largest series of multicentric giant cell tumours was reported by Hoch *et al*. from the Mayo Clinic.[6] They reported 30 cases among which 19 were of metachronous lesion locating mostly around knee joint. Three cases of metachronous ulnar involvement were reported in which the second lesions were in the same bone. In this case, the ulnar tumour was removed by wide margin excision but the metachronous lesion appeared in ipsilateral distal radius after an interval of 2 years. It was neither a missed lesion nor recurrence as it was completely absent in all previous imagings including bone scans and MRI and then developed purely in another bone with origin in distal radius metaphysis rather than cortex.

After adequate resection of the tumour, reconstruction of osseous defect is necessary to preserve the function and alignment. One option was to accept a one bone forearm with ability of flexion and extension retained at elbow but no supination and pronation. The authors reconstructed the ulnar osseous defect with free vascularised osteocutaneous fibular graft restoring all ROM and alignment.

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