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## Two rare cases of Giant cell tumor of Distal Ulna

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## ABSTRACT

Giant cell tumours, though benign, are locally aggressive bone tumours with a relatively high recurrence rate. These usually occur in distal radius, distal femur, proximal tibia and humerus. Treatment options for contained lesions at these sites include joint preservation procedures such as extended curettage with cementing or bone graft. GCT in spine, calcaneum and distal ulna are rare, with no uniform consensus regarding the ideal treatment.

Here we report two cases of GCT distal ulna managed with extended curettage and polymethylmethacrylate cementing showing good functional and radiological outcomes without signs of recurrence during 2 years follow up.

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## 1. Introduction

Giant Cell Tumour (GCT) accounts for 3–5% of all bone tumours. They are usually benign with local tissue spread and high recurrence rates. Metastases may occur in 1%–9% of patients with GCT and some earlier studies correlate the incidence of metastasis with aggressive growth and local recurrence [1]. They are common among females in the 3rd to 5th decades. Despite low mortality rates, GCT cause significant morbidity in peri-articular locations due to distortion of the local bony architecture leading to stiffness and decreased function of the adjacent joint.

The distal epiphysis of the ulna is an unusual place for a primary bone GCT; in fact, this occurs in only 0.45%–3.2% of all primary bone GCTs [2].

## 1.1. Case 1

A 19 year old female with complaints of pain in the wrist since 3 months was evaluated clinically and radiologically. On clinical examination, a swelling measuring 4 cm × 3 cm × 2 cm was found in the distal aspect of ulna along the subcutaneous border. Skin over the swelling was smooth and pinchable. There was full range of movement at the wrist with no distal neurovascular deficits. Plain radiographs showed a lytic lesion in the meta-epiphysis of ulna 3 cm × 2 cm × 2 cm **Radiograph 1** MRI showed a T1- hypo and T2- hyperintense lesion in the subarticular region of ulna with cortical break in the antero-medial aspect with no articular extension.



Fig. 1. Bluish-brown lesion with thinned out cortex.

No periosteal thickening or matrix calcification was seen. Tru-cut biopsy confirmed Giant-cell tumor. Intraoperatively a brownish mass with a hemorrhagic, cystic component was noted. Extended curettage with bone cementation was done (**Figs. 1–3**).

Postoperative period was uneventful. The patient had complete functional recovery (**Fig. 4**) by 3 months and was followed up for 2 years with no signs of recurrence clinically or radiographically (**Radiograph 2–3**).

## 1.2. Case 2

A 31 year old female presented with history of progressively increasing swelling along the medial aspect of right distal forearm for 4 months. On examination, there was pain on deep palpation over the swelling. There was no restriction of range of movements. Plain radiograph taken at the district hospital showed a lytic lesion in the distal ulna (**Radiograph 4**). She was referred to Medical Col-

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**Radiograph 1.** Lateral and AP views showing lytic lesion in the distal ulnar with thinned out medial cortex not involving articular surface.



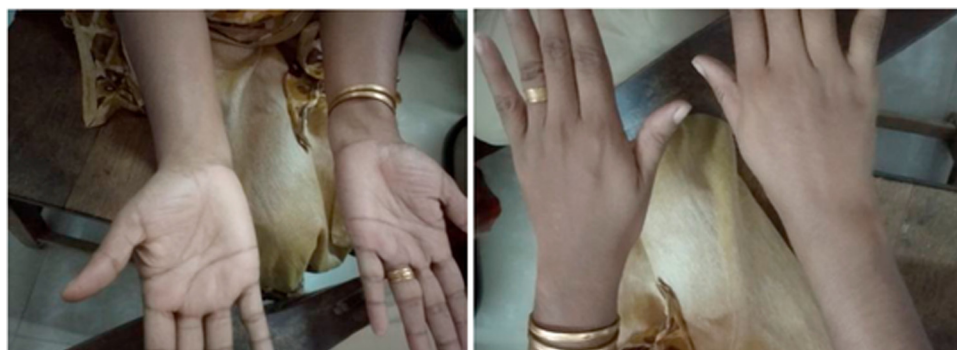
**Fig. 2.** Extended curettage of the distal ulna.



**Fig. 3.** Defect filled with PMMA cement.

lege Hospital Kozhikode for further evaluation. MRI showed a lesion measuring 3cm × 1cm × 2 cm in distal ulna not extending to the articular surface with no cortical break. There was no periosteal reaction or calcification or soft tissue extension. Tru-cut biopsy showed giant cells interspersed with normal bone. Extended curettage of the lesion was done after making a window in the lateral wall and filled with PMMA bone cement (**Radiograph 5**). Histopathology showed low grade spindle cell tumor with giant cells in abundance with clear margins confirmatory of Giant cell tumor. Post-operative period was uneventful. On 2 year follow-up

patient is doing well with no signs of local recurrence clinically or radiologically (**Fig. 5, Radiograph 6**).



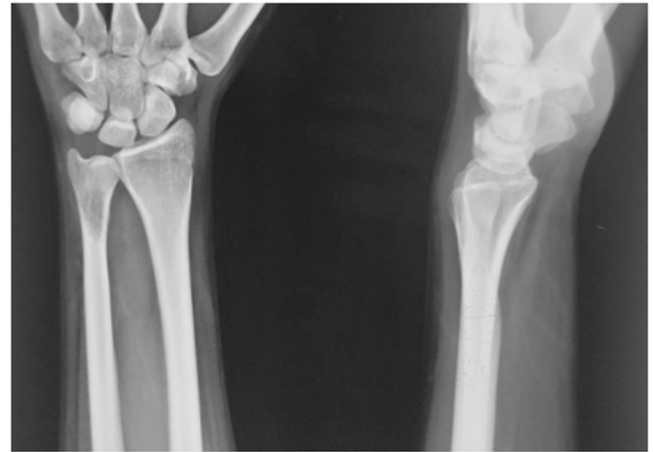
**Fig. 4.** Complete functional recovery at 3 months postop with full pronation and supination.



**Radiograph 2.** 1 year follow-up.



**Radiograph 3.** 2 Year follow-up shows no recurrence.



**Radiograph 4.** Shows lytic lesion with geographic margins in distal third of ulna.

**2. Methods**

The case report was structured according to the submission guidelines of the journal and reported according to the Surgical case report(SCARE) protocol [3].

**3. Discussion**

Giant cell tumors are locally aggressive tumors of the epiphysis found in skeletally mature patients with predilection for sites like distal end of radius, proximal humerus, distal femur and proximal tibia.

**4. Radiological characteristics**

Plain radiography is the first investigation advised by an orthopaedist or a referring clinician for the diagnosis and assessment of a suspected GCT. In locations like pelvis and spine they may be obscured due to overlap by normal bone and might be missed in initial stages. This leads to delay in diagnosis. The radiological features of a classical GCT includes an eccentric lytic lesion in the epiphysis with well-defined (mostly geographical) margins devoid of sclerosis with thinning of cortex. Wide zone of transition, soft tissue extension, thinning and destruction of cortex indicate local aggression.

Aneurysmal bone cyst (ABC), which is a radiological differential can be identified by occurrence in middle age, metaphyseal location, ballooning of cortex with intra-lesional loculations and absence of periosteal reaction, unless associated with pathological fracture. Magnetic resonance Imaging (MRI) is useful in subarticular lesions to exclude joint affliction and to map the metaphyseal and soft tissue extension (tendon, synovium and neurovascular structures) as the management in these cases change drastically. Occult cortical breaks, especially those along the distal radio-ulnar joint and interosseous border of radius can be better detected on an MRI.

**5. Treatment**

Treatment strategies vary from simple curettage to wide excision and joint sacrificing procedures. Irrespective of the treatment modality used, the following principles are adhered to:

1. Proper planning and preoperative assessment of the lesion.
2. Histopathological confirmation of diagnosis before any definitive procedure.





**Radiograph 5.** Post-op day 2 xray AP and Lateral.



**Fig. 5.** Full functional outcome at 3months post-op.

3. Achieving adequate local control with appropriate surgical methods and adjuvants.
4. Preservation of available joint function without compromising on degree of tumour resection.
5. Prevention of local recurrence and metastasis.

Currently available surgical options include:

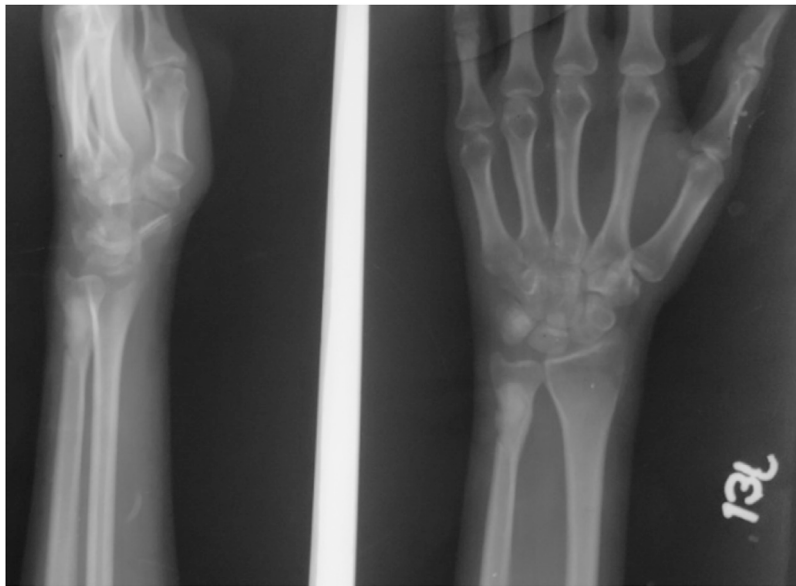
- Intralesional curettage
- Extended Curettage and bone grafting
- Resection followed by allograft reconstruction
- En bloc resection with or without reconstruction

- Embolization of the feeding vessels

Adjuvants used to ablate tumour cells include:

1. Cryosurgery with liquid nitrogen
2. Phenol/Ethanol/Hydrogen peroxide
3. Polymethymethacrylate (PMMA) bone cement
4. Argon laser

Treatment outcomes are determined by tumour-related characteristics including size, location, biological activity, cortical bone destruction or evidence of pathologic fracture [4]. En bloc resection



**Radiograph 6.** Post-op 2 years; No signs of recurrence or articular collapse.

is a radical procedure eradicating the tumour and reducing the risk of recurrence but shows poor functional outcome.

In the past few decades, local control with preservation of joint function has been attempted by intralesional curettage and corticocancellous bone grafting from iliac crest or tibia. Intralesional excision has a high recurrence rate (60%) on account of the residual tumour cells invariably left behind. Adequate exposure of the lesion is the sine qua non to ensuring complete tumour removal [5]. Creation of a large cortical window without overhanging shelves provides the best access for curettage. Ridges are broken using high powered burrs. Small pockets might be identified with adequate light and accessed using multi-angled curette [6]. Any remaining tumour cells are washed out using a pulsatile jet-lavage system and burring of the remaining cancellous bone. [7–9]. Bone cementation is an alternative for dead space management, especially in lower limb GCTs which require structural support for weight bearing. The recurrence rate after cementing is 5–8% [10,11]. This is in contradiction to the findings of the Canadian Sarcoma Group study [12] which reported a 17% overall recurrence rate irrespective of filling material or the type of adjuvant. According to some studies, adjuvants are unnecessary in cases such as intraosseous GCT [13]. Others state that curettage alone provides inadequate tumour clearance and adjuvants provide better results globally, especially in terms of functionality. The key is to attain a balance between complete oncological clearance and skeletal segment functionality [13–15]. Presentation with a pathologic fracture (12%) often heralds more aggressive disease, higher recurrence rates and metastatic spread [16–20]. Such cases may warrant a more radical surgical debridement and reconstruction.

## 6. Polymethylmethacrylate (PMMA) bone cement

Positive results have been obtained by using polymethylmethacrylate (PMMA) bone cementing of the defect [21]. Several theories regarding mechanism of action have been proposed.

1. Local hyperthermia due to the exothermic reaction of PMMA which induces necrosis limited to neoplastic cells [22].
2. Chemical cytotoxicity produced due to PMMA polymerization.

Adjuvants like Methotrexate, Adriamycin and other cytotoxic agents are also incorporated in bone cement. Rock et al. reported a reduced recurrence rate (10%) after adjuvant treatment with cement, vis-à-vis curettage alone (10–47%) [23].

However, the effects on major weight-bearing joints, cartilage and subsequent risk of arthritis due to replacement of subchondral region with cement remain controversial [24,25]. A suggested method to avert the risk of late articular degeneration is the “sandwich” multilayer reconstruction technique [26]. Subarticular lesions with less than 1 cm of residual subchondral bone after extended curettage are suitable for implementing this method. A mixture of morcellized corticocancellous autograft (approximately 1 cm thick) is packed beneath the subchondral bone. A layer of gelfoam is layered over this and the remaining space is packed with cement. Thermal damage from the curing cement is prevented. This method also confers the benefit of pre-empting damage to articular cartilage during cement removal in case of recurrence. There are reports that support use of topical or systemic bisphosphonates as adjuvant therapy for GCT. They limit the tumour progression by inducing apoptosis of osteoclast-like giant cells [27–31].

## 7. Conclusion

GCT of Distal Ulna is a rare entity. Extended curettage followed by PMMA cementing is an effective treatment with low recurrence even in subarticular GCTs.

## Declaration of Competing Interest

The authors report no declarations of interest.

## Funding

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## Ethical approval

No ethical committee approval was needed for this case report.

## Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

## Author contribution

**Dr Puneeth K Pai:** Conceptualization, Methodology, Software Data curation, Writing- Original draft preparation. Visualization, Investigation, Software, Writing Reviewing and Editing, Validation.

**Dr Jeejesh Kumar T K:** Supervision.

**Dr Priyavrata Rajasubramanya:** Editing.

## Registration of research studies

N/A.

## Guarantor

Dr Puneeth K Pai.

## Provenance and peer review

Not commissioned, externally peer-reviewed.

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