International Journal of Orthopaedics Sciences

E-ISSN: 2395-1958
P-ISSN: 2706-6630
IJOS 2021; 7(4): 114-118

www.orthopaper.com
Received: 19-08-2021
Accepted: 21-09-2021

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Management of distal ulna giant cell tumor by en bloc resection and reconstructed by a novel technique

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DOI: https://doi.org/10.22271/ortho.2021.v7.i4b.2873

Abstract
Giant cell tumor (GCT) is a locally aggressive benign primary bone tumor accounting for 20% of all the benign primary bone tumors. Peak incidence is between 20 and 45 years with slight female predominance. Distal end of ulna is a rare site for any primary bone tumor, especially 2-4 for giant cell tumors with reported incidence of 0.45 to 3.2% in literature. Considering local aggressive nature and high recurrence, resection is the treatment recommended. In such cases, instability of the ulnar stump and ulnar translation of the carpals are known 3-6 complications causing reduced grip strength. Literature review states, reconstruction with iliac bone graft and stabilization of the ulnar stump with tenodesis using extensor carpi ulnaris. We report a 34 year old female with giant cell tumor of the distal ulna treated by en bloc resection and reconstruction of the distal radio-ulnar joint (DRUJ) and triangular fibrocartilage (TFCC) complex with proximal fibula, palmaris longus graft with augmentation by extensor carpi ulnaris tenodesis and stabilization of the proximal stump with dynamic compression plate. This newer technique of DRUJ and TFCC reconstruction with proximal fibula and palmaris longus graft following en bloc resection of the distal ulna had excellent functional results with no evidence of recurrence after seven months follow-up. This case is presented for its rarity and novelty of the technique used for the anatomical reconstruction.

Keywords: Giant cell tumor, distal ulna, en bloc resection, proximal fibular graft, palmaris longus graft

Introduction
Giant cell tumor (GCT) or Osteoclastoma is a rare, benign tumor with local aggressive nature. It accounts for 4-5% of all primary bone tumors, commonly affecting distal femur, a proximal tibia, distal radius and proximal humerus. Pulmonary metastases are seen in 2% of patients with GCT. Local recurrence rate is approximately 25% with various types of intraluminal ablation procedures described for the GCT. En bloc resection of the GCT is the recommended treatment option with fewer chances of recurrence at this location. We report a patient with GCT of the distal ulna, treated by en bloc resection, followed by reconstruction of the distal ulna and triangular fibrocartilage complex (TFCC) using proximal fibula, palmaris longus tendon and a part of extensor carpi ulnaris with stabilization of the proximal stump using dynamic compression plate. To the best of our knowledge, this is a novel technique to be reported.

Case report
A 34-year-old South Indian female, presented with a single painless swelling of left distal ulna of one and half years duration. Initially the ulna head was more prominent compared to the opposite side. It gradually increased to present size. There was no history of fever, loss of appetite or weight and there was no other significant history. On examination, the swelling was oval in shape measuring 5.0 x 4.0 cm, occupying the distal third ulna (Fig.1 & 2) Skin over the swelling was normal in colour and temperature. The swelling was diffusely tender, firm in consistency and was fixed to the underlying bone. But skin over the swelling was pinchable. The range of movements of the wrist was full and painless. Hand grip was normal in both hands with normal distal neurovascular status. There was no lymphadenopathy or any other systemic disease. Hematological and biochemical parameters were within normal limits. Plain chest radiograph was normal.

Plain radiograph of the left wrist in both AP and lateral views showed single, expansile, multiloculated lytic lesion involving the epiphysis and metaphysis of distal end of ulna with no cortical breach or periosteal reaction (Fig.3). It extends upto the subchondral bone of articular cartilage with no intralesional calcifications. CT scan showed expansile lytic lesion with cortical thinning and areas of cortical destruction. (Fig.4) MRI both plain & contrast showed 6.5 x 5.6 x 5 cm, isointense T1W and hyperintense T2W lesion in the distal ulna (Fig.5,a & b) involving the epiphyseal-metaphyseal region which enhances with contrast. Cortical break was noted with TFCC involvement. Also edema of the adjacent tissues was present. Both clinically and radiologically the features were suggestive of giant cell tumor of the distal end of ulna.

Fig. 1: Clinical picture of the dorsum of the Lt.wrist

Fig. 2: Clinical picture of the dorsum of the Lt.wrist

Fig. 3: X-ray of Lt. forearm & wrist in both AP & Lat views showing an expansile, multiloculated lytic lesion of distal ulna

Fig. 4: CT image of the Lt. forearm & wrist in both axial (A) & coronal (B) section shows an expansile, lytic lesion of distal ulna with cortical thinning and area of cortical disruption

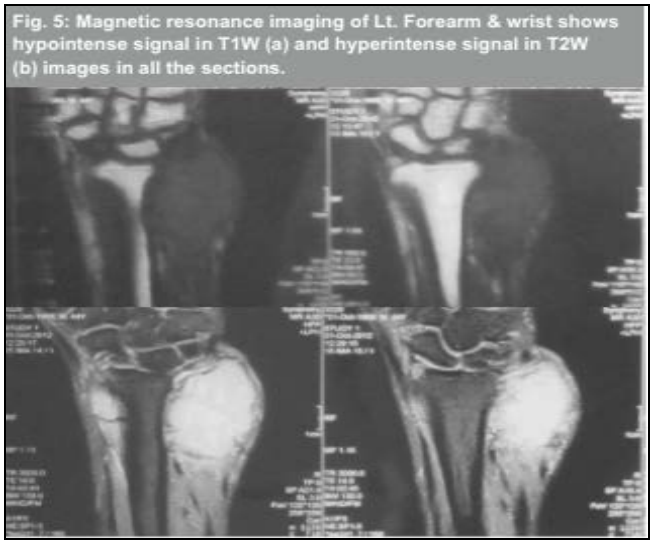

Fig. 5: Magnetic resonance imaging of Lt. Forearm & wrist shows hypointense signal in T1W (a) and hyperintense signal in T2W (b) images in all the sections.

Fine needle aspiration cytology showed a double cell population consisting of stromal cell clusters and numerous osteoclastic giant cells. This confirmed our diagnosis as a giant cell tumor of distal ulna. Based on the clinical and radiological evaluation, the tumor was graded as aggressive type (grade 3) as per the enneking classification of giant cell tumors of bone. Hence, as per the recommended treatment, en bloc resection was planned. The disease, prognosis with chances for metastasis or recurrence and treatment advised were all discussed with the patient in detail and informed written consent was obtained.

The patient was taken up for extra periosteal en bloc resection of the tumor. Through a 10 cm incision over the dorsal and medial aspect of the left distal ulna, the tumor was exposed completely. The extensor carpi ulnaris (ECU) tendon was found almost engulfed by the tumor mass (Fig.6a). Sparing the ECU, the tumor was resected en bloc with 2cm cuff of normal bone (Fig.6b). The excised specimen was 8 cm (6+2 cm) in length and 4 cm in breadth (Fig.6c).

Fig. 6: Intraoperative clinical picture showing GCT of Lt. Dstal ulna almost engulfing the ECU tendon (a), enbloc resection of GCT with 2 cm cuff of normal bone (b), resected specimen (c)

Considering the age of the patient and high level of functional demands, distal ulna with distal radio-ulnar joint (DRUJ) and triangular fibrocartilage complex (TFCC) reconstruction was planned. Then a proximal fibula of 9 cm was harvested and the fibular head was resized to match the size of the ulnar head (Fig.7a). After preparation, the proximal fibular graft was kept in the defect, with resized fibular head in place of ulnar head. Precaution was taken to prevent the raw area of the resized fibula from facing the radius, this is to prevent cross union at DRUJ. Palmaris longus tendon was harvested and used as a tendon graft for the reconstruction of the TFCC and DRUJ as described by Adams and Berger in the treatment of chronic DRUJ instability (Fig.7b). This construct was stabilized with two parallel k-wires across the fibular graft and distal radius (Fig.7c). Also ECU tenodesis was done to augment the construct (Fig.8a & 8b).

Fig. 7: Intraoperative clinical picture showing resizing of fibular head (a), reconstruction of the DRUJ using Palmaris longus graft (b), reconstructed distal ulna and stabilised DRUJ (c).

Fig. 8: Intraoperative clinical picture of the reconstructed distal ulna and DRUJ after augmentation with ECU (a,b), immediate post operative radiograph of Lt. Forearm and wrist (c).

Proximally the fibular graft was stabilized with the proximal ulnar stump using a 6 holed dynamic compression plate. The k-wires were kept outside the skin for easy and earlier removal of the same. Wound was closed in layers with drain left in situ. Left upper limb was immobilized in above elbow (AE) cast with elbow in 90 deg flexion and forearm in full supination (Fig.8c). Wound inspection and suture removal was done through a window made in the cast. After wound healing, AE cast with elbow in 90 deg flexion and forearm in full supination was reinforced and continued till 6 weeks. K-wires were removed at 6 weeks. After that, gentle elbow, forearm, wrist and hand mobilization exercises were started.

Fig. 9: Clinical picture of the Lt. Forearm, wrist and hand after six months (a) and radiograph of Lt. Forearm and wrist (b) at twelve months follow-up.

Follow-up was advised at 3 months, 6 months and once in six months for two years. After one year, sound union of the graft was noted with good functional range of movements at wrist and forearm with excellent hand grip and no evidence of carpal instability or recurrence of the tumor.

Discussion

Primary GCT of distal ulna is rare, with reported annual 2, 3, 4 incidence of 0.45 to 3.2%. Various treatment options are available based on the clinical and radiological staging of tumors by Enneking. Stage I or Latent lesions require no active treatment. Stage II or Active lesions require intralesional curettage with adjuvant therapy. Stage III or Aggressive lesions require en bloc resection. Cooney *et al*, achieved excellent results in 75% of their eight cases, which required wide excision of the distal ulna for various tumors. He concluded that routine reconstruction of the osseous defect is not needed for all cases. On the other hand, many authors have documented, poor functional results following en bloc resection of the distal ulna for those reasons like pain and restriction of movements due to impingement of the ulnar stump on the radial metaphysis and dorsal winging of the ulnar stump or radio-carpal instability with ulnar translation of the carpus. The distal ulna was initially considered as a dispensable bone and distal ulnar resection was recommended for degenerative conditions as Darrach's procedure. Though distal ulnar resection following any tumor is not comparable to Darrach's procedure for various reasons the later procedure too has unpredictable long-term results. Hence, there arises the need for anatomical reconstruction of the distal ulna and the distal radio-ulnar joint with stabilization of the proximal ulnar stump for better functional results especially in young patients with high functional demands. Various authors suggested different techniques for reconstruction or stabilization after en bloc resection of distal ulna for primary neoplasms. These include stabilization by tenodesis, auto graft or allograft replacement, plate arthrodesis and bone transport using ilizarov ring fixator. Gainer reported lasso tenodesis of the ulnar stump with tendon graft from Palmaris longus, in two cases of GCT after 15 wide excision. Ferracini *et al* reported five patients with GCT and 3 patients with other tumors involving distal ulna. They stabilized ulnar stump using flexor carpi ulnaris (FCU), fascia lata and autograft. They suggested that soft tissue stabilization of the ulnar stump should be done whenever possible. Goldner and Hayes in 1979, was the first to describe ECU tenodesis to stabilize the ulnar stump based on their experience on 225 patients, who underwent distal ulna excision for various conditions. Following them, many the authors reported this procedure after wide excision of the distal ulna. Wurapa and Whipple reported a two stage allograft reconstruction of the DRUJ. With 40 months follow-up. patient had extremely satisfactory results.

Stofflen *et al*. reported the use of ilizarov fixator for the transport of proximal ulnar stumps. Patient had an acceptable and painless range of movements of the wrist. Hashizume *et al*. described an ulnar buttress arthroplasty following en bloc resection of the distal ulna for the first time. They used a longer iliac bone graft on the ulnar side of the radius using a screw and k-wire to attain the buttress against axial loading. They reported excellent results with six months follow-up. Similarly, Minami *et al*. Reported satisfactory results by

following a technique of modified ulnar arthroplasty in a young adult after resection of GCT of the distal ulna. Monappa A Naik *et al*. reported excellent functional outcome with their technique of ulnar buttress arthroplasty, almost similar to minami *et al* report, following en bloc resection of GCT distal ulna in a 34 year old female. From the literature review, we can understand that, reconstruction of DRUJ along with stabilization of the ulnar stump is mandatory in any functionally demanding individual, following en bloc resection of the distal ulna. In our technique, instead of iliac crest graft, proximal fibula is used for the reason of more anatomical similarity to the distal ulna. We had replaced the fibular graft, for the entire length of the ulna resected. Proximally a six holed narrow DCP was used to stabilize the fibular graft with the ulnar stump. Also in our technique, DRUJ & TFCC was reconstructed with Palmaris longus graft using the technique, similar to the one described by Adams and Berger for DRUJ reconstruction in chronic 20 DRUJ instability. Hence, there is no question of either ulnar stump instability or radio-carpal instability with translation of the carpals in our technique. In literature, ECU tenodesis was done only to stabilize the proximal ulnar stump. But In our technique, ECU tenodesis was done for the augmentation of the DRUJ and not for the stabilization of the proximal ulnar stump.

Conclusion

Our case is to be reported, using this entirely novel technique of reconstruction of the entire length of excised distal ulna, DRUJ and TFCC using proximal fibula, Palmaris tendon graft, ECU, K-wires and dynamic compression plate for GCT of distal ulna in a 34-years-old functionally demanding female with excellent functional results in range of movements, stability and good hand grip with no evidence of recurrence after one year follow-up.

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