



Is there a need for reconstruction after excision of the distal ulna for Giant-Cell tumour ?

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Giant cell tumour rarely involves the distal ulna ; the literature has sporadic cases reported, and the need for reconstruction after distal ulnar resection remains controversial. The uncommon presentation often leads to diagnostic delays, and some of the cases reported have attained large sizes prior to diagnosis, leading to procedures which range from soft tissue stabilization to bone graft reconstruction of the surgical defect. We analyzed our cases with specific reference to the need for additional reconstruction ; the radiological and functional outcome of extra-periosteal distal ulna resection without reconstruction in 4 patients with giant cell tumour of the distal ulna was assessed at a mean follow-up of 9 years post surgical excision. In our experience there was no radiological evidence of recurrence, radioulnar convergence and ulnar translocation of the carpus. Functional outcome was graded as excellent in 3 and good in one patient. In conclusion, it could be suggested that there is no need for reconstruction after distal ulna resection for GCT as an adequate functional outcome is seen even in late diagnosed cases. Reconstructive procedures, which come at the cost of additional morbidity and complications, and requires special technical skills, are not routinely justified for this rare condition.

Keywords : giant cell tumour ; distal ulna ; distal ulna resection ; distal ulna reconstruction.

INTRODUCTION

Giant cell tumour (GCT) is one of the most common bone tumours, accounting for 20 % of biopsies

confirmed benign lesions (7). It usually affects young adults aged 18 to 40 years, with a slight female preponderance (7). The tendency of GCT to recur locally, develop benign pulmonary secondaries, or even become frankly malignant has caused considerable debate and confusion over the benign or aggressive nature of this tumour, as well as confusing the options available for management (20). The distal femur and the proximal tibia are the most common sites and account for approximately 60% of cases (13,31). Involvement of the distal ulna is rare, with a reported incidence varying from 0.5% to 2.9% of all GCTs (3,7,10,15,18,27,29,30).

The golden rule of GCT management is complete tumour eradication (oncological control) with maximum preservation of adjacent joint function. Enneking stage 1 and stage 2 GCTs can be managed by intralesional resection (extended curettage), but recurrence rates approaching 17% have

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been reported (5,12,31). Wide resection is usually reserved for Enneking stage 3 GCTs (12,31,33) ; this however invariably jeopardizes the adjacent joint function.

Giant cell tumours of the distal radius or ulna have been described as difficult to treat, chiefly because of their proximity to carpal bones (1,9,18). In underdeveloped countries like India, the problem is compounded by late presentation, when the tumour has expanded to a fairly large size. At this stage, en bloc resection is the treatment of choice, but the operative approach is tempered by the fact that potential problems could occur with regard to the instability at the distal radioulnar joint (DRUJ). The more commonly involved distal radius has been addressed adequately in the reported literature, with well defined procedures describing reconstruction of defects created by resection of the distal radius either by arthrodesis or proximal fibular interposition (8,12,23). Distal ulnar resections, however have been much less discussed, principally due the fact that they are uncommon presentations and thus less experience is recorded. Even the literature is confusing, with reports advocating no reconstruction after resection (10,35), to soft tissue stabilization procedures (14,21), to bone graft augmentation (19,36). Resection with no subsequent reconstruction may have consequences such as a painful wrist, weakened grip and difficulties on pronation and supination due to radioulnar convergence (2,4,16,17). Due to the lack of consensus in the published literature on this specific issue, we evaluated our cases, and present our experience.

MATERIALS AND METHOD

In a retrospective review of our records from 1990 to 2004 at the Post Graduate Institute of Medical Education and Research, Chandigarh, India, we found 7 cases diagnosed with GCT of the distal ulna. Four patients with complete records were available for long term follow-up and review. Serum calcium and phosphorus levels were normal in all the patients, ruling out any underlying parathyroid disease. On reviewing the patient records of the three patients who were lost to follow-up, all were females and two of them had an involvement of the non-dominant (left) hand. These three patients had a stage 3 GCT according to Enneking staging and were treated by

en bloc extra-periosteal resection. Mean age at presentation in these three patients was 26 years.

All cases were assessed with special reference to initial clinical presentation, duration of symptoms, time between initial diagnosis and surgical intervention, and type of ultimate definitive management. The Enneking system of GCT classification was followed (13). Previous radiographs and other imaging modalities were studied, and surgical notes were analyzed for procedural details.

En bloc extra-periosteal resection was done in all four patients to achieve a wide margin. Extra-periosteal resection included the excision of the DRUJ capsule, triangular fibrocartilage complex and origin of pronator quadratus along with the tumour. The level of resection of the ulna was determined by the extent of bony involvement as seen on the radiographs. Histopathological examination confirmed the diagnosis of GCT in all the patients. Special note was made of level of resection and post operative complications.

At follow-up (Jan-Feb 2009), all four patients were assessed for functional and oncological outcome. They were examined for five parameters ; namely wrist pain, radiocarpal joint stability, ulnar stump stability, range of motion at the wrist joint and grip strength. Functional results were graded as excellent, good, fair and poor using the grading scheme developed by Cooney *et al* (10). Radiocarpal joint stability was assessed by determining the radio-ulnar and volar-dorsal translation of the hand and carpus relative to the stabilized distal end of the radius, and findings were compared with the normal contralateral side. Ulnar stump stability was assessed by movements of the stump relative to the stabilized distal radius.

Oncological outcome was assessed by clinical and radiological examination for any evidence of recurrence at the time of follow-up. Follow-up radiographs were also assessed for any features of radioulnar convergence (2).

RESULTS

Three female and one male patient were available for the long term follow-up. Their mean age at presentation was 27.5 years (25 to 36 years). The non-dominant limb was involved in 3 patients. Average follow-up was 9 years (range 3 to 17 years). Demographics of the patients are detailed in table I.

All patients had presented with slowly growing swellings for 2 to 3 months in the distal forearm.

Table I. — Patient Demographics

	Age/sex	Side	Profession	Enneking staging	Ulnar length resected	Follow-up period
Case 1	37 yr/F	Non dominant	Staff nurse	Stage 3	6.5 cm	17 yrs
Case 2	25 yr/F	Non dominant	House wife	Stage 2	5 cm	9 yrs
Case 3	26 yr/F	Dominant	Stenographer	Stage 3	8 cm	7 yrs
Case 4	30 yr/M	Non dominant	Mechanic	Stage 3	9 cm	3 yrs



Fig. 1a,b,c,d. — Pre-operative radiographs of GCT of the distal ulna in 4 patients. a) A huge osteolytic lesion of distal ulna without any cortical margins. b) well-contained expansile lesion with sclerotic margins and septations. c) osteolytic lesion of distal ulna with a pathological fracture. d) recurrence of GCT with ill-defined margins and a pathological fracture.

Treatment at that stage had varied from massage by quacks to minor surgical procedures by qualified doctors, and recurrence had occurred locally in one female patient. The swelling was painful in 3 patients, while the fourth patient had presented with a fairly large but painless swelling of the distal ulna.

Radiographic record review showed the presence of well defined osteolytic expansile lesions in the distal ulna, abutting the subchondral plate (fig 1a,b,c,d). One was a huge osteolytic lesion with no cortical margins visible, one was a contained expansile lesion with sclerotic margins and septations; one expansile lesion had presented with pathological fracture, and one recurrence after

inadequate surgery had an angulated pathological fracture. MRI (available in our institute for the latter cases) was available in the last case (fig 2); the lesion was noted to be expansile and destructive. CT scan had been done in the case with recurrence (fig 3).

On evaluating Enneking staging, 3 patients had stage 3 GCT with associated soft tissue component and 1 had a stage 2 tumour. Extraperiosteal resection (fig 4) was done in all the patients; the length of distal ulnar resection ranged from 5 to 9 cm in the four patients. No reconstruction was done for the osseous defect created in all the patients. The recorded oncological results were excellent, and at follow-up we had no clinical or radiological signs



Fig. 2. — T2 weighted image of the distal forearm showing hypotense signal changes in the distal ulna GCT with the soft tissue extension.



Fig. 3. — CT scan image of the distal ulna in the patient with recurrence showing complete erosion of the cortical margins.

of recurrence (fig 5). In addition, there was no radiological evidence of radio-ulnar convergence and ulnar translocation of the carpus.

Functional outcome was excellent in 3 (fig 6) and good in one patient, who had prominence of the distal ulnar stump, but no functional impairment. None of our patients had to change their profession. No patient complained of any pain or restriction of movements of the wrist in activities of daily living. All patients had a stable wrist while doing heavy work, and radiocarpal stability was not hampered. Grip strength, measured with a dynamometer, was reduced minimally in three of our patients as compared to the contralateral side; this was not considered subjectively significant by our patients. The ulnar stump was prominent in one case but there were no clicking while doing movements of the wrist.

There was an average terminal restriction of 8° and 12° in wrist flexion and wrist extension respectively as compared to the contralateral normal side. Supination of the forearm, however, was comparable to the contralateral side, but there was terminal restriction of pronation (average 5°); ulnar and

radial deviation was also restricted by an average of 7° .

DISCUSSION

The incidence of distal ulna GCT varies in different reported studies. Harness *et al* (18) reported an incidence as low as 0.5%, while Campanacci (7) reported a 1.2% and Blackley (3) a 2.9% incidence. The overall low incidence of GCT at this site is reflected in table II, which is a review of large series of GCT reported in the last 40 years. These studies reported 41 cases of GCT of the distal ulna out of a total of 3222 cases of GCT, with a cumulative incidence of 1.27% (3,7,10,15,18,27,29,30). Even at our institution, we came across 7 cases of GCT involving the distal ulna out of 528 overall cases of GCT, which is an incidence of only 1.3%. A Medline search with the keywords Distal ulna- Giant cell tumour came up with only 60 hits, many of which were cases with distal radius GCT. The above



Fig. 4. — Intra-operative photograph of the extraperiosteal resection of distal ulna demonstrating the origin of the pronator quadratus muscle.

discussion highlights the rarity of GCT involving the distal ulna and correspondingly management options are also vaguely defined for tumours in this area. Series reporting patients with primary distal ulna GCTs are rare, but many case reports exist with proponents of excision alone (10,35), soft tissue stabilization (14,21) and excision with bone graft (19,36). Since all of these additional procedures add to the morbidity and complication rates, and require varying levels of additional technical skill and instrumentation; we proposed to analyse if these additional procedure were justified or not.

The distal end of the ulna contributes significantly to the stability of the wrist for a good grip in the hand because of its relationship with the distal radius, carpus and TFCC. So, there is possibly a theoretical disadvantage in the form of ulnar translocation of carpal bones, if the distal ulna is excised. The insertion of the pronator quadratus is another important anatomical structure, which can cause problems after distal ulna excision. Resection at a lower level could potentially cause impingement symptoms due to the pull of this muscle, while resection at a higher level causes instability and undue prominence of the ulnar stump. Even the Darrach procedure was criticized for the ulnar stump instability and radio-ulnar convergence leading to pain and clicking during rotatory movements of the wrist (2) in spite of its excellent functional results by achieving painless supination and pronation in patients with DRUJ dysfunction (6,11,24,25, 26). Many stabilizing procedures such as tenodesis (2,11), pronator quadratus muscle transfer (28), combination of the two (22), ulnar lengthening osteotomy (34) and ulnar head prosthesis (32) were recommended to avoid these complications. The functional outcome following en bloc resection of the distal ulna without reconstruction has been reported in two studies. Cooney *et al* (10) have studied the functional outcome in patients with 3 malignant and 5 benign neoplasms of the distal ulna at a mean follow-up of 7.5 years. Wolfe *et al* (35) have



Fig. 5 a,b,c,d. — Follow-up radiographs of 4 patients showing no evidence of recurrence, radio-ulnar convergence and ulnar translocation of carpus.



Fig. 6. — 17 years follow-up clinical photographs of a patient (case 1) showing adequate functional outcome and range of motion of the wrist joint.

Table II. — Literature review : GCT of distal ulna

Study	Year	Total cases of GCT	Cases of GCT distal ulna	Incidence of distal ulnar GCT
Goldenberg <i>et al</i> (15)	1970	218	4	1.83%
Reddy <i>et al</i> (27)	1974	108	1	0.92%
Sung <i>et al</i> (30)	1982	208	3	1.44%
Dahlin <i>et al</i> (27)	1986	429	8	1.86%
• Campanicci <i>et al</i> (7)	1987	327	4	1.22%
• Schajowicz <i>et al</i> (29)	1991	465	4	0.86%
Memorial Sloan-Kettering Cancer Centre records (10)	1997	265	3	1.1%
• Blackley <i>et al</i> (3)	1999	135	4	2.9%
• Harness <i>et al</i> (18)	2004	539	3	0.5%
Present study	2009	528	7	1.3%
Total		3222	41	1.27%

studied the results of distal ulnar resection in 9 patients with pain following DRUJ excision and 3 patients with neoplastic conditions. Both of these studies had concluded that excision of a substantial portion of the distal ulna can be performed without concern for subsequent instability, ulnar translocation of the carpus and radioulnar convergence.

All patients were also assessed for the various criticisms made for Darrach's procedure in previous studies (2,4,16,17). None of our patients complained of painful clicking on movements of the wrist and there was no associated pain on compression of radius and ulna. Follow-up radiographs did not show scalloping of the ulnar border of the distal radius, which also ruled out radiological evidence of radio-ulnar convergence. So, there were no features of impingement in all our patients even at

the longest follow-up of 17 years. There were no complaints regarding instability at the wrist joint while hooking up heavy objects. One of the patients (fig 6) continues to work as an ICU Nurse in our institution 17 years after resection.

The unique feature of our study was the size of the distal ulnar GCTs which we encountered. This was attributed to the delayed presentation in conjunction with the aggressive nature of the tumours, or perhaps it might have something to do with the local site (which may not be conducive for intra osseous growth), and cortical expansion occurring at an early stage. Three out of four tumours were classified as Enneking stage 3 and one as Enneking stage 2 GCT. When the distal ulna is resected for tumour excision, oncological aspects like adequate margins take precedence over anatomical and

functional aspects to prevent the recurrences. Oncological outcome was excellent in all of our patients and there was no recurrence even at the mean follow-up of 9 years. Functional outcome was excellent in three and good in one of our patients. None complained of ulnar snapping or painful clicking on movements of the wrist, which is an important feature of radio-ulnar convergence, except one patient who had a prominent ulnar stump without any functional impairment. The variation in resection level in our 4 cases had no bearing on the ultimate functional outcome. All patients had good grip strength, no significant carpal translocation as evident on the follow-up radiographs (fig 5). As discussed in literature, the satisfactory functional outcome after en bloc resection of the distal ulna could be due to the intact central band of interosseous membrane which plays an important role in the stability of the remaining ulna (35).

In conclusion, distal ulnar resection should be the first line of management for aggressive giant-cell tumours of the distal ulna. None of our patients had to change their profession postoperatively. As all of our patients had a painless and stable wrist with intact functional range of movement and no painful ulnar snapping, it can be suggested that there is no need of reconstruction of the defect created by extraperiosteal distal ulnar resections for aggressive giant cell tumours.

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