Limb Salvage Surgery in Large and Fungating Giant Cell Tumors of Distal Radius: A Case Series

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INTRODUCTION

A Giant Cell Tumor (GCT) is a benign tumor characterized by aggressive behavior and recurrence tendency or metastasis. GCT represents five percent of all primary bone and mostly occurred in the distal radius, distal femur, proximal tibia, humerus, and sacrum. Ninety percent of GCT is encountered in the epiphyseal region [1]. Zou et al. reported that 48.3% of GCT presented in small dimensions with a mean size of 4.7 cm. In the series of 58 patients with GCT, it was found that tumors with a diameter of larger than 5 cm had a 3.893 times higher rate of recurrence [2].

GCT treatment aims to complete the resection of the tumor to lower the chance of recurrence and to preserve as much wrist function as possible. Various surgical technique was considered for GCT of distal radius treatment, from intra-lesional curettage to wide or en-bloc resection, followed by reconstruction, arthrodesis, ulnar translocation, bone graft, spacer or prosthesis, and amputation [1].

In the case of recurrence of GCT distal radius, en-bloc resection is commonly adopted to achieve adequate margin [2]. However, more destructive lesion requires complex surgical reconstruction with the consequence of a higher complication rate and may result in amputation [2,3]. Complications following en-bloc resection include wrist joint dislocation and subluxation, autograft fracture, non-union, infection, injury of a major artery, and sensory disturbance or motoric palsy [2]. Infection is one of the common causes of amputation [3].

Case series reporting limb salvage on a large and fungating distal radius GCT is very limited. The treatment of large and fungating distal radius GCT increases the risk of complications including amputation; hence, it requires more complex surgery and reconstruction than usual GCT. We need to explore more choices for the treatment or information regarding the characteristic...
of this kind of tumor. In this case series, we prefer to perform the limb salvage surgery with wide en-bloc resection, followed by reconstruction of the bone and soft tissue, instead of amputation.

CASE PRESENTATION

We reported four cases of large and fungating GCT at the distal radius. Patients were all young adults (age range 27 to 46 years old) with large GCT Campanacci grade III on the wrist. The demographic characteristics of the cases were presented in Table 1.

Table 1. Characteristics of the patients

<table>
<thead>
<tr>
<th>Case</th>
<th>Gender</th>
<th>Age (years)</th>
<th>Size of Tumors</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Male</td>
<td>27</td>
<td>12 cm x 8 cm x 6 cm</td>
</tr>
<tr>
<td>2</td>
<td>Female</td>
<td>31</td>
<td>10 cm x 10 cm x 8 cm</td>
</tr>
<tr>
<td>3</td>
<td>Female</td>
<td>32</td>
<td>11 cm x 9 cm x 9 cm</td>
</tr>
<tr>
<td>4</td>
<td>Female</td>
<td>46</td>
<td>17 cm x 14 cm x 8 cm</td>
</tr>
</tbody>
</table>

In case 1, a 27-year-old male with pain and enlarging lump on his left wrist. It occurred for 9 months before hospital admission. Physical examination revealed a 12 cm x 8 cm x 6 cm mass with a fungating mass on the radial aspect of the distal radius on the left wrist (Figure 1a). The mass was solid and immobile, with a smooth surface, radial nerve palsy, and hypoesthesia. There was a limited range of motion (ROM) on the wrist and fingers joint due to lump and pain.

In case 2, a 31-year-old female with a 1-year history of pain and swelling on her left wrist. The mass has been rapidly growing in the past 2 months. A solid, immobile, smooth surface with a 10 cm x 10 cm x 8 cm size was found with soft tissue destruction (Figure 1b).

In case 3, a 32-year-old female with a history of enlarged right wrist mass since 8 months ago. An 11 cm x 9 cm x 9 cm size mass with fungating wound and venectation was visible. Radiography showed a radiolucent lytic in the metaphyseal region of the distal radius (Figure 1c). In case 4, a 46-year-old female with gradual onset of pain and swelling of the right wrist for 8 months. There was a 17 cm x 14 cm x 8 cm solid mass over the right distal radius with tenderness on palpation, venectation, and shiny skin. Distal sensibility was normal compared to the healthy one (Figure 1d).

All the patients were examined by forearm X-ray and MRI depicted a lytic lesion with a geographic pattern at the left distal metaphyseal region of radial with cortical breakage on expanding mass and ballooning with soft tissue involvement. A histopathological result from biopsy shows round-oval-spindle, single core tumor cell proliferation with diffuse growth forming fasciculus, confirming GCT of bone. The patients were diagnosed with GCT on the left distal radius Campanacci grade III.

Surgical method and characteristics of the tumor.

These four distal radii GCT cases underwent limb salvage. The procedure consists of wide resection, bone and joint reconstruction, and soft tissue management. Dorsal-proximal forearm approach was carried out by passing the biopsy site, ulcer, fungating wound, and other compromised skin, then directing towards volar on the distal side and towards dorsal again to get better exposure of radial artery as well as a superficial branch of the radial nerve. The radial arteries were preserved (Figure 2a), except for case number 4, in which the artery has encaged within the tumor, so we ligate the artery. After the tumor was exposed (Figure 2b), we carefully separated it from the tendon, blood vessels, and nerve to protect them. Tendons of abductor pollicis longus, extensor pollicis brevis, extensor indicis propius,
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Figure 2. Surgical procedure. (a) Preservation of tendons, nerve, and blood vessels in case 1; (b) Tumor mass exposed in case 2; (c) Tendons and vessels were pushed aside to facilitate tumor removal in case 3.

Figure 3. The defect filled with bone cement spacer after tumor removal.

and 2nd and 3rd digit of extensor digitorum communis were found to be embedded within the tumor. The aforementioned tendons were dissected and debrided to remove any remaining malignant tissue (Figure 2c).

The wide resection of the bone is done by osteotomy 2 cm from the margin of healthy bone. After the tumor was resected, the site was debrided by saline and ethanol 96% as adjuvant local therapy. Bone cement was inserted as a spacer to the defect and to reduce the risk of local recurrence (Figure 3). Bone reconstruction continued with fixation by intramedullary K-wire, then the joint was temporarily fixed by the ulna-carpal joint wire and polyethylene terephthalate suture. Dissected tendons were repaired and sutured. Subsequent soft tissue defect was managed by groin flap on cases 1, 3, and 4 and primary suture for patient number 2 (Figure 4). After the surgery, post-operative X-ray imaging was taken to ensure the position of the bone cement spacer and K-wire (Figure 5). Second-stage surgery was then performed to release the flap, together with shoulder, elbow, and finger joint manipulation under general anesthesia. The biopsy sample was taken at the tumor margin along with healthy tissue to ensure the tumor site has been freed from malignant cells.

All cases have similar characteristics regarding their extension. They grew more dorsally rather than toward the anterior aspect, infiltrating extensor tendons; especially in compartments 1, 2, and 3. The superficial branch of the radial nerve was infiltrated in all four cases. The radial artery was shoved away by the tumor and no ulnar artery was found to be involved. The mass extends to the ulnar bone, which caused partial cortex erosion. The volar extension is relatively smaller than the dorsal and always encapsulated with expanding pronator quadratus muscle.

Short-term follow-up was done with an average of 9 months; pain reduction was measured by VAS score before the surgery and the latest time of follow-up; upper extremity function measurement using the quick Disabilities of the Arm, Shoulder, and Hand (DASH) score; complications, also the recurrence and metastases were measured. The complications were divided into minor...
quick DASH score was 50.0, 34.1, and 52.3 for patients 1, 2, and 4. No data is available for patient no. 3. All the patients had a minor complication, that is, the disturbance of superficial radial nerve distributions. No patient had any major complications, local recurrence, or metastases. Post-surgery biopsy for all of the cases showed no atypical cells representing malignancy. The summary of tumor size, pain reduction, quick DASH score, complication, and recurrence or metastasis were presented in Table 2.

and major. The criteria for minor complications were using are a superficial infection that does not require debridement, mild sensory disturbance, dislocated or subluxation joint, and implant or cement break. While for the major complications are deep infection requiring debridement, debilitating pain or complex regional pain syndrome, major vascular or compromised nerve, and other conditions that lead to amputation.

All patients had reduced scale of pain as measured with VAS (visual analog scale) score. The post-operative DASH score was 50.0, 34.1, and 52.3 for patients 1, 2, and 4. No data is available for patient no. 3. All the patients had a minor complication, that is, the disturbance of superficial radial nerve distributions. No patient had any major complications, local recurrence, or metastases. Post-surgery biopsy for all of the cases showed no atypical cells representing malignancy. The summary of tumor size, pain reduction, quick DASH score, complication, and recurrence or metastasis were presented in Table 2.

Figure 4. Inguinal flap was performed and left for 2–3 weeks.

Figure 5. Pre and post operative imaging in case 1.

Table 2. Patient characteristics by tumor size, post-operative pain, quick DASH score, complications (major and minor), and recurrence or metastases.

<table>
<thead>
<tr>
<th>Patient</th>
<th>Size (in cm)</th>
<th>Pain Reduction</th>
<th>Quick DASH Score</th>
<th>Complications</th>
<th>Recurrence or metastases</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>VAS before VAS After</td>
<td>50.0</td>
<td>Minor</td>
<td>Major</td>
</tr>
<tr>
<td>Patient 1</td>
<td>12x8x6</td>
<td>6 2</td>
<td>50.0</td>
<td>+</td>
<td>-</td>
</tr>
<tr>
<td>Patient 2</td>
<td>10x10x8</td>
<td>7 1</td>
<td>34.1</td>
<td>+</td>
<td>-</td>
</tr>
<tr>
<td>Patient 3</td>
<td>11x9x9</td>
<td>6 2</td>
<td>NA</td>
<td>+</td>
<td>-</td>
</tr>
<tr>
<td>Patient 4</td>
<td>17x14x8</td>
<td>7 2</td>
<td>52.3</td>
<td>+</td>
<td>-</td>
</tr>
</tbody>
</table>

DASH: Disabilities of the arm, shoulder, and hand questionnaire; VAS: Visual analogue scale
DISCUSSION

A giant cell tumor of bone is a benign tumor but tends to be locally aggressive with relatively high local recurrence [6]. Distal radius GCT as well as the proximal femur has been shown to have a higher local recurrence than in other sites [7]. Ten percent of cases are located at the distal radius [6]. It was reported that distal radius GCT could exhibit larger sites and be more aggressive than usual [5]. Tumor sites of ≥ 5 cm in diameter had a 3.893 times higher rate of recurrence [2].

The treatment of distal radius GCT aims for complete tumor resection, lowers the risk of recurrence, and preserves wrist function. Treatment options include amputation, ulnar translocation with arthrodesis, curettage, bone grafting, curettage with bone cement filling, and en-bloc resection of the lesion with reconstruction [6]. Amputation is the most common method performed in managing large GCT tumors due to its progressive and destructive nature [5]. However, since GCT is considered not a true malignancy with a low rate of recurrence and metastasis, there is a higher chance to preserve the anatomy and functional ability. Hence, limb salvage surgery should be considered a promising option to manage large GCT.

There are limited pieces of literature reporting on limb salvage procedures in fungating tumors of bone. In patients with large tumor presentation, as seen in Figure 1, some surgeons prefer to do the amputation. Parsons et al reported about 35% of cases with fungating lesions are managed with amputation [4]. Rangaswamy et al. reported limb salvage surgery in fungating GCTs and one of the cases was a radius GCT. They performed arthrodesis with a plate to fuse the wrist joint, but eventually ended up with amputation due to surgical site infection [5]. Ng et al. reported a study of 31 GCT patients, 3 of them had amputation which included 1 patient with GCT of the distal radius with lung metastases [8]. Takeuchi also reported 2 of 103 patients with recurrent GCT were amputated [9]. Shresha et al. reported 1 case of fungating GCT of the distal ulna, with a 10 x 10 cm wide tumor mass, and managed with wide resection and fusion of the wrist joint [10]. In our study, two patients (numbers 1 and 3) have fungating mass, while other patients have compromised skin with fungating tendency. We managed to salvage the limb and avoid complications leading to amputation.

Wang reported a significant difference in VAS score on pre-and post-operative patients with radius GCT treated with osteoarticular allograft and 3D printed prosthesis (5.5 (range, 4–7) preoperatively and 1.3 (range, 0–4) postoperatively (P < 0.01) [11]. Zou et al showed an average VAS score of 2.5 ± 0.9 in a patient treated with intralesional curettage [2]. In our study, the average VAS Score was decreased, which indicate that any choice of therapy resulted in a lower VAS score and showed good result.

The Size of GCT of Bone (diameter ≥ 5 cm vs. 5 cm HR: 3.893, 95% CI: 1.109 to 13.659, p = 0.034) are the two independent risk factors related to local relapse; in a study by Zou et al, intralesional curettage group achieved much better functional scores and much fewer complications (non-unions, dislocations, fractures, and infections) compared to resection ones [2]. In our study, the mean site of the tumor was all the patients who were treated with resection and all of the cases showed no major complications. The mean size of the tumor was 12.5 x 10.25 x 7.75 cm.

A study by Singh et al. showed the functional outcome of the application of non-vascularized fibular graft following distal radius GCT excision in 15 patients with an average DASH score of 25.2 in a patient without soft tissue compromise [12]. Ramadhany et al. reported the functional outcome after wrist arthrodesis following excision of the distal radius is moderate disability according to the DASH score on 3 patients [13]. Zou et al. reported that in GCT treated with intralesional curettage the average DASH score was 9.1 ± 3.9 with the average diameter of the tumor was 4.7 ± 1.3 cm [2]. DASH score in our study is still acceptable, with a mean value of 45.5. It suggests that smaller size, tumor with minimal soft tissue involvement results in better functional outcome.

There are various anatomical restrictions in treating a distal radius giant cell lesion. It is closely associated with the radio-carpal and the radio-ulnar joints, also the muscle cover is relatively limited. In addition, various important vessels, nerves, and tendons need to be protected to preserve optimum hand function. All these factors need to be taken into consideration when treating distal radius GCT to strike the right balance between complete disease clearance and retaining good hand function. At the same time, the recurrence and complication rates need to be minimized.

Intra-lesional curettage had a high local recurrence rate but it is useful in preserving wrist movements, functions, and stability. The use of fibular autograft gives more congruency of the carpal joint, rapid incorporation, and absent immunogenic reactions. The use of fibular vascularize autograft will take 12 – 14 hours of operation time and will sacrifice two major vessels [6]. In our case series, we did not perform any bone grafting procedure because of the high possibility of recurrences in large or contaminated distal radius GCT. It is found that the use of 3D printed cementless prosthesis compared with allograft had a better outcome in early-stage application but had a similar outcome on functions and complications [16]. Aggressive curettage with a high-speed burr had a lower recurrence rate than simple curettage [7]. Our treatments were done using wide
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resection to ensure low local recurrence and bone cement insertion with screw and plate fixation to preserve the limb.

A study by Panchwagh et al. reported that the recurrence rate of distal radius GCT is about 32% in 24 patients and 36% of those cases are classified as Campanacci grade III cases [14]. Blackley et al. stated that despite the high rates of recurrence reported in the literature after treatment of GCT with curettage and bone grafting, it suggests the risk of local recurrence after curettage with a high-speed burr and reconstruction with autogenous +/- allograft bone is similar to that observed after the use of cement and other adjuvant treatment [15].

Malu et al. did a limb-preserving procedure in GCT at the distal radius with en-bloc resection and reconstruction arthroplasty with non-vascularized proximal fibular autograft. 18 cm long dorsal ‘S-shaped’ incisions were made 3 cm from the distal wrist joint. Radical en-bloc of tumor resected and with 4 cm of the distal radius, leaving 14 cm bone defect. Reconstruction was done using an ipsilateral proximal fibular non-vascularized graft of 8 mm more than the actual length to prevent subluxation. The articular surface of the fibular head was placed over the scapholunate articular surface and fixed to the carpals with K-wires. The fibular graft was fixed with a dynamic compression plate. The newly created fibula-ulnar joint was stabilized with another K-wire [6]. In a case where GCT was found after ORIF treatment for a wrist fracture in a pregnant woman, the use of a high-speed burr, removal of the cortical surface, bone cement placement, and fixation with a plate and two screws shows no recurrence or other complications two years postoperation [17]. In the cases presented before, it also shows that the patients had no pain, no complications, and normal biopsy post-surgery. The exothermic reaction that occurs while the cement cures are also required to increase the tumoricidal effect. GCT is relatively recurrent in 4 years, furthermore, with large and fungating GCT, we need to reduce the recurrence rate and might need the second stage operation. In our study, we use 96% alcohol and cautery also we use bone cement as a spacer to decrease the recurrence.

Large and fungating tumors have a high risk for local recurrence, so we prefer to use bone cement as a spacer instead of bone autograft, to avoid waste morbidity of the graft, especially at the donor site. Bone cement also has an advantage as it acts as a local adjuvant therapy effect on the tumor to reduce the risk of recurrence [18]. Zekry et al recommended that a bone cement spacer is indicated for a defect in the upper limb. Filling the intramedullary cavity of the bone with bone cement or antibiotic-containing bone cement may improve mechanical strength. In addition, bone cement has several advantages such as better screw hold and pull-out strength of the screws fixed to bone cement [19].

This study used a groin flap for the reconstruction of the soft tissue defect in three patients. We understand that the groin flap was not ideal to close the defect. We did not use the local flap like the radial forearm flap because the distal artery was still compromised, and we did not sacrifice more arteries where the radial forearm flap performs. Before taking the flap, we also took a tissue sample proximal to the tumor border to ensure the tissue is free from the tumor, so we can be sure the defect is safely closed with the groin flap.

Since three decades ago, pedicled flaps from the groin and abdomen have been the workhorse for reconstruction in the hand and forearm. However, these flaps have several disadvantages such as stiffness which may cause patient discomfort and risk of contamination in the groin area. In this era of microsurgery, free flaps started to replace pedicled flaps. Groin flap is still indicated in several cases such as larger or multiple defects within the hand or forearm, similar to this case [20].

Previous research reported local adjuvant treatment after curettage is unnecessary, but some recommended the use of adjuvant treatment to reduce local recurrence. Treatment with curettage and phenol has a similar local recurrence rate with curettage and ethanol. Alcohol 96% is thought to have an adjuvant therapeutic effect. The administration of denosumab was presumed to have a good effect on GCT treatment, but recent studies show that it did not decrease local recurrence [7]. In a study by Tsukamoto et al., it was even found that patients treated with denosumab after curettage still had a higher local recurrence rate [21]. Chemotherapy is also required for pain reduction and tumor suppression. However, due to its complexity, major arteries and nerves could be affected [22]. In our study, we did not give chemotherapy or denosumab. The limitation of this study is the short follow-up time, including one patient who came at 2 months post-operative only, so the author still needs a longer follow-up to see if there is complication or metastases.

CONCLUSIONS

This case series showed that the treatment of large and fungating GCT of the distal radius with limb salvage procedure consists of resection, bone and joint reconstruction, and soft tissue management for soft tissue defect and still can give the satisfying result in VAS Score, Quick DASH Score, and major or minor complication; thus, giving options other than amputation. Longer follow-up on this study is still required for giving a better result.
DECLARATIONS

Competing interest
The authors declare that they have no competing interests.

Ethics approval and consent to participate
The patient agreed to publish his/her case to increase awareness among residents, physicians, surgeons, and medical staff.

Acknowledgment
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REFERENCES