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ORIGINAL ARTICLE

Functional outcome of en bloc excision and custom prosthetic replacement for giant cell tumor of the distal radius

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Abstract

Background Giant cell tumors (GCT) of the distal radius at Campanacci grade II/III are particularly challenging to treat. Wide excision is the management of choice, but this creates a defect at the distal end of radius. We treated 11 cases of GCT of the distal radius by en bloc excision and custom prosthetic replacement. The purpose of this study was to present our experience and assess the functional outcomes of all patients treated with this surgery.

Materials and methods Between 2005 and 2014, we followed up 11 patients with GCT of the distal radius who were treated with en bloc excision and custom prosthetic replacement. All cases were evaluated based on clinical and radiological examinations, passive range of motion (ROM) of the wrist joint, complications, and Musculoskeletal Tumor Society (MSTS) score.

Results Mean follow-up period was 55.5 months (24–83 months); mean resected length of the radius was 7.9 cm. One patient had tumor recurrence in the soft tissues after 15 months (recurrence rate 9.09 %). No patient had fracture, recurrence in the bone, metastases, or immune rejection. No complications were seen, such as loosening, rupture, or dislocation of the custom prosthesis. One patient developed superficial infection at the operative site which resolved after a course of antibiotics for 4 weeks. One patient experienced pain, which could be endured without the need for analgesics. Average ROM was 40.9° of dorsiflexion, 30.0° of volar flexion, 46.4° of supination, and 38.2° of pronation. Mean grip strength was 71 % (42–86 %). Overall revised MSTS score averaged 80.3 % (63.3–93.3 %) with one being excellent, five good, and five satisfactory.

Conclusion En bloc excision and custom prosthetic replacement for a Campanacci grade II/III GCT of the distal radius results in reasonable functional outcome at intermediate follow-up evaluation. Although average ROM of the ipsilateral wrist is poorer than some studies with other techniques, this method can be considered a reasonable option.

Introduction

The distal radius is a relatively common skeletal site for primary bone tumors, and ~10 % of all giant cell tumors (GCT) occur in the distal radius, which is the third most common location (after the distal femur and proximal tibia) of GCT [1]. Because of the frequency of recurrence in Campanacci grade II/III GCT [2] after curettage with adjuvant inactivations, such as high-speed drill, bone cement, electric cauterization, liquid nitrogen, and alcohol, en bloc resection that completely removes the tumor and reconstruction following tumor resection in this location might be necessary in aggressive cases, which exhibit extraosseous extension or recurrence after previous treatment [3–5].

Although providing the best chance of cure from GCT, en bloc excision of the distal radius poses a challenging problem of skeletal reconstruction and functional restoration because of the high functional demands of the hand, the limited surrounding soft tissue, and the proximity of important nerves and tendons. Various techniques, including resection arthroplasty [6], use of a nonvascularized or vascularized autogenous fibular graft [7–10] and allograft

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replacement [11–13], prosthetic replacement [14–16], ulnar translocation [18], and arthrodesis [19] have been used for reconstruction. These techniques all come with their unique possible advantages and complications, but a gold standard for distal wrist reconstruction has not been established.

Despite extensive experience with prosthetic replacement of juxta-articular tumors of the lower limb, there are few reports of prosthetic replacement of the distal radius [14–17]. We reviewed 11 patients with GCT of the distal radius managed by en bloc resection and custom prosthetic replacement, which was chosen by the patient. The purpose of this study was to assess medium outcomes of functional results, complications, and oncological outcomes of patients who underwent this procedure.

Materials and methods

On retrospective search of our hospital records, 11 patients with distal radial tumors who underwent en bloc resection and custom prosthetic replacement between 2005 and 2014 were analyzed in this study. Six patients had primary GCT, and five had recurrent GCT after tumor curettage and bone grafting. There were six men and five women, with a mean age of 33.7 years (range 26–46 years). Four patients had right-sided surgeries and seven had left-sided surgeries. All patients who had a primary lesion had a needle biopsy for diagnosis before the operation. Preoperative investigation of each patient included: radiographs of the forearm, wrist, and chest; computed tomography (CT) scan; magnetic resonance imaging (MRI); and emission computed tomography (ECT) bone scan of the lesion when appropriate. According to Campanacci's radiological grading method [20], two patients were grade II and nine were grade III.

Prosthesis

An individual custom-made prosthesis of the distal radius with appropriate dimensions obtained from preoperative radiographs (lidakang Technology, Beijing, China) was used in all cases. Based on the accumulation of experience, the design has been modified and upgraded over the years. The basic components include a high molecular weight polyethylene liner and distal radial shaft components in titanium (Ti) alloy, comprising a proximal stem, body, and cushion base (Fig. 1). The articular surface of the polyethylene liner is designed concave to fix the prosthesis to carpal bones. The prosthesis body replaced the distal radius defect after tumor resection, and the proximal stem was fixed with cement to the radial shaft. Multiple pores in the prosthesis, to be conducive with soft tissue reconstruction, are created, if necessary.

Fig. 1 Distal radius custom prosthesis, including a high molecular weight polyethylene liner and distal radial shaft composed of proximal stem, body, and cushion base

Surgical technique and postoperative management

Patients were operated under general anesthesia, and the ipsilateral leg and arm were prepped and draped appropriately. Through a standard dorsal approach to the wrist and including the previous biopsy site and operating incision, en bloc resection of the tumor was performed. Bone was resected at a level determined preoperatively based on the extent of bone involvement on plain X-rays and MRI, plus a safe margin of 2–3 cm. On average, 7.9 cm (6–11 cm) of bone was resected. Dissection was extraperiosteal in order to avoid spillage of tumorous tissue, and a soft tissue cuff was excised along with the tumor, taking care not to damage neurovascular structures. During the operation, we found that in all patients, the lesion did not penetrate the articular cartilage, thus allowing preservation of carpal bones. We attempted to avoid resecting all radiocarpal ligaments, tendons, and joint capsule if they were not involved, as these were later repaired through pores attached to distal radial prosthesis, forming a stable wrist joint. A custom-made distal radial prosthesis with appropriate dimensions obtained from preoperative radiographs was used for reconstruction, which was inserted into the proximal radius using cement. Maintaining the length of the radius is paramount for balance of ligaments and tendons. The cut ligaments, wrist capsule, and fibrocartilage complex were sutured to the prosthesis. After careful hemostasis, the wound was closed over a suction drain. Postoperatively, the limb was immobilized in an above-elbow cast for 4 weeks, then active range of motion (ROM) wrist exercises were allowed and gradually increased in intensity depending on patient

tolerance and progress. All patients were advised not to engage in contact sports or strenuous activities.

Evaluation of clinical outcomes

After institutional review board approval, patients were contacted by letter and/or telephone and invited to participate in the study. We attained informed consent and routinely scheduled visits at 1, 3, 6, 9, 12, and 18 months after surgery and yearly thereafter until last follow-up. During the latest scheduled follow-up, nine patients came to our hospital for physical and radiographic examinations.

Two patients visited a hand surgeon in their local community and posted the corresponding data to us. We evaluated the outcomes clinically and radiographically for tumor recurrence, infection, loosening, rupture, custom prosthesis dislocation, and other complications. Forearm and wrist ROM were assessed, and grip strength was measured and compared with the contralateral wrist using a grip-strength meter. At the most recent follow-up, functional results were analyzed using the revised Musculoskeletal Tumor Society (MSTS) score, which rates patients based on factors pertinent to the patient as a whole (pain, functional activities, emotional acceptance) and those specific to either upper limb (hand position, manual dexterity, lifting ability) [21]. Results were established as excellent for MSTS score >90 %, good for 80–90 %, satisfactory for 60–80 %, and poor for ≤60 %. All patients were evaluated during the final follow-up by an independent research fellow blinded to primary surgical treatment.

Results

The study group consisted of 11 patients; one patient (patient 4) was Campanacci grade II, with pathologic fractures. The mean resected radius length was 7.9 cm (range 6–10 cm). No patient had tumor recurrence in the bone; however, one (patient 11) experienced recurrence (recurrence rate 9.09 %) in soft tissues 15 months later, which was subsequently resected, and no recurrence was seen at follow-up.

Follow-up time ranged from 24 to 83 (average 55.5) months. ROM and grip strength were compared with the normal contralateral side (Table 1). Functional results revealed that the average active ROM of the wrist was 40.9° (range 20°–60°) of dorsiflexion, 30.0 (range 15°–45°) of volar flexion, 46.4° (range 20°–65°) of supination, and 38.2° (range 10°–60°) of pronation; mean grip strength was 71 % (42–86 %). Movement and finger and thumb sensation were satisfactory in all patients. Overall revised MSTS score averaged 80.3 % (63.3–93.3 %), with one being excellent, five good, and five satisfactory. Detailed data on clinical results are reported in Table 1. Preoperative and 54-months' follow-up radiographs of patient 5 are given in Figs. 2 and 3 and functional photographs in Figs. 4 and 5.

There were no major complications: One patient developed superficial infection at the operative site, which resolved after a 4-week course of antibiotics. One patient experienced pain (especially upon dorsiflexion), but the pain could be endured without the need for analgesics, and no sign of arthritis was observed at the radiocarpal joint. No patient had fracture, recurrence in the bone, metastases,

Table 1 Clinical results of follow-up evaluations of 11 patients

Patient number	Age (years)/ gender	Campanacci grade	Follow-up (months)	Range of movement (ROM)				Grip strength in mmHg (% of contralateral side)	MSTS score[a]
				Flexion (°)	Extension (°)	Supination (°)	Pronation (°)		
1	32/F	III	83	25	20	25	10	14 (44)	63.3 (19)
2	27/M	III	30	40	60	65	60	30 (86)	93.3 (28)
3	46/M	III	24	15	25	35	30	16 (52)	76.7 (23)
4	27/M	II	80	30	30	50	45	27 (84)	83.3 (25)
5	26/F	III	54	45	50	60	55	24 (83)	86.7 (26)
6	34/F	III	79	35	50	65	60	20 (76)	90.0 (27)
7	36/M	III	39	30	35	45	30	25 (73)	76.7 (23)
8	29/M	III	77	35	55	55	45	26 (77)	86.7 (26)
9	42/F	III	55	15	20	20	15	12 (48)	66.7 (20)
10	40/M	II	41	35	60	65	50	26 (75)	83.3 (25)
11	32/F	III	49	25	45	25	20	15 (53)	76.7 (23)
Mean	33.7		55.5	30.0	40.9	46.4	38.2	22.6 (68.3)	80.3 (24.1)

[a] Musculoskeletal Tumor Society (MSTS) score calculated as percentage of maximum possible score of 30

Fig. 2 Preoperative X-ray (anteroposterior and lateral views) of patient 5 showing Campanacci grade III giant cell tumor (GCT) of the distal radius
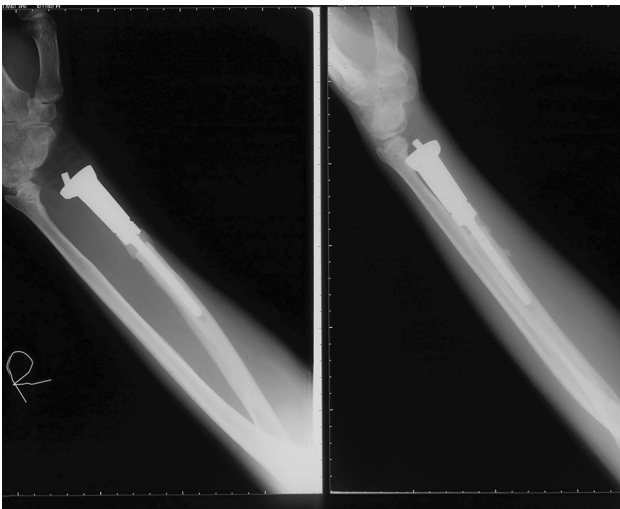

Fig. 3 Postoperative X-ray of patient 5 after 54 months (anteroposterior and lateral views)

or immune rejection, and there were no complications such as loosening, rupture, or prosthesis dislocation.

Discussion

The clinical behavior of GCT is unrelated to histological grading [22], and the decision to either salvage or excise tumorous bone is based on the ability to achieve stability and function by whatever means possible [23]. Indications for en bloc resection would thus include pathological fractures, extensive bone involvement with large soft tissue involvement, and articular surface collapse [23, 24]. For Campanacci grade III, Campanacci grade II

Fig. 4 Functional photograph of patient 5 after 54 months (dorsal flexion)

Fig. 5 Functional photograph of patient 5 after 54 months (palmar flexion)

with pathological fractures, and recurrent GCT of the distal radius, lesion curettage is not feasible due to a lack of residual bone stock and radiocarpal joint disruption. Furthermore, Campanacci grade III tumor recurrence rates have been reported to be as high as 70 % with curettage

alone [25]. Therefore, patients in our study were highly suitable for en bloc resection.

Reconstruction of defects that remain after GCT excision of the distal radius presents a substantial challenge in orthopedic oncology. Achieving adequate tumor clearance without the risk of local recurrence while preserving good hand function proves to be a daunting task due to the unique anatomical constraints, such as proximity to complex joints, limited soft tissue cover, and the close relationship to important vessels, nerves, and tendons. There is no consensus in the limited literature about the best surgical reconstruction of the distal radius after tumor resection; although multiple methods are described in small patient populations [6–19]. However, a standard procedure has not yet been formulated as a result of the small numbers of patients, differences in surgical techniques, and patient characteristics.

Because of anatomical similarities between distal articulation of the radius and the proximal aspect of the fibula, reconstruction using nonvascularized or vascularized fibular grafts with and without arthrodesis has thus far been the method of choice for distal radius reconstruction. Advantages of these reconstructions include preservation of motion by earlier graft incorporation and hypertrophy. Although outcomes of such reconstructions are promising, complications are not uncommon, including nonunion, delayed union, graft fracture, wrist-joint subluxation, and donor-site morbidity. Patients from four series who underwent fibular graft reconstruction [7–10] showed limited wrist ROM and improper articulation leading to progressive and accelerated degenerative changes of the carpofibular joint. Additionally, this reconstruction technique is a technically demanding surgery with prolonged operating time, two major vessels need to be sacrificed, and extensive radiographic studies of the vascular pattern of limbs are necessary.

Osteoarticular allograft reconstruction is another common method, which has the advantages of no donor-site morbidity, shorter operating time, and greater radiocarpal joint congruency. Some researchers [11–13, 26, 27] reported that allograft implantation is an excellent option for reconstruction in patients with an aggressive tumor, but some complications, including nonunion, fractures, slow incorporation of the allograft, wrist osteoarthritis, and possibility infection transmission, have been observed for distal radius allograft implantation after osteoarticular resection. Although a painless, stable, and functional reconstruction of the wrist could be achieved using an osteoarticular allograft, this reconstruction has been preferred for smaller forearm resections with possible preservation of the wrist extensors. Moreover, lack of allograft availability and specialized bone-bank facilities in most countries may prevent its frequent use.

Translocation of the ulna is an effective reconstruction method but may not provide cosmetically acceptable results, as there is narrowing of the wrist and distal forearm, giving the limb an hourglass appearance [18, 28]. Radiocarpal arthrodesis using either osteoarticular bone allograft or tumor prosthesis may result in a successful salvage procedure after failed joint reconstruction. An arthrodesis offers inherent stability, pain relief, adequate hand function, and good grip strength, despite being at the cost of wrist mobility. An additional advantage of wrist arthrodesis is the possibility of bearing substantial loads [29]. Other authors critically commented on the use of an arthrodesis because it was associated with fairly high rates of graft fracture, donor-site morbidity, and limited wrist function [19].

Endoprosthetic replacement of the distal radius has also been attempted by some authors (Table 2), which has several advantages, including function preservation function, anatomy restoration, and the ability to repair large defects while avoiding delayed union and donor-site morbidity. Initial attempts to replace the distal radius with a prosthesis have failed, necessitating arthrodesis or amputation [14, 15]. However, Hatano H et al. [17] presented two cases in 20060 of massive bone defects of the distal radius in which alumina ceramic prosthetic replacements were used. The authors evaluated patients >10 years after the procedure; both patients had degenerative changes to the wrist, and both had returned to their previous occupation after surgery. So, the authors believed that reconstruction using a ceramic prosthesis was a reasonable alternative to using autograft for the patient with a massive defect of the distal radius. This method results in little postoperative pain, a moderate ROM, and satisfactory function. Natarajan et al. [16] reported results of 24 cases of aggressive benign and malignant tumors of the distal radius treated by resection and prosthetic replacement with a custom megaprosthesis with a bipolar hinge manufactured locally. Average follow-up was 78 months, average MSTS functional score was 75 %, and the 10-year prosthesis survival rate was 87.5 %. The authors also believed that custom prosthetic replacement for malignant and aggressive benign tumors of the distal radius had proven to produce good functional outcomes with an acceptable complication rate (infection, skin-flap necrosis, aseptic loosening). Our results are comparable with the studies reported by Hatano et al. [17] and Natarajan et al. [16], with fewer complications and better functional results.

However, results of such procedures have not been conclusively shown to be better than existing treatments, as most reports are either case reports or very small series [7–17]. Previous studies and this current study used different types of prosthesis in terms of design and materials. For example, prostheses comprised acrylic with a long stem

Table 2 Prosthetic replacement for giant cell tumor (GCT) in the distal radius in the English literature

Authors	Number	Reconstruction	Follow-up (months)	recurrence rate (%)	Complications	Failed	Flexion/extension	Supination/pronation	MSTS score[a]	Grip strength in mmHg (% of contralateral side)
Gold [14]	1	Distal radial prosthesis	59	0	None	0	NA	NA	NA	Sufficient for manual work at 59 months
Gold [15]	1	Distal radial prosthesis	144	100	Fracture of the prosthesis, increasing pain and disability	1 (amputaion)	NA	NA	NA	NA
Hatano et al [17]	2	Distal radial ceramic prosthesis	168, 120	0	None	0	15°/30°, 15°/35°	45°/30°, 50°/60°	83, 83	22 (71), 29 (85)
Natarajan et al [16]	24	Bipolar hinge custom megaprosthesis	78 (range 24–156)	0	Skin-flap necrosis (2), infection (2), aseptic loosening (2)	2 (wrist arthrodesis)	25° (range 5°–40°)/20° (range 10°–50°)	40° (range 5°–80°)/60° (range 25°–85°)	75	NA
This study	11	Custom-made distal radial prosthesis	55.5 (range 24–83)	9.09	Superficial infection (1), pain (1)	0	30° (range 15–45)/40.9° (range 20–60°)	46.4° (range 20–65°)/38.2° (range 10°–60°)	80 (range 63.3–93.3)	22.6 (68.3 range 44–86)

NA not applicable

[a] Musculoskeletal Tumor Society (MSTS) score calculated as percentage of maximum possible score of 30

of stainless steel [14], alumina ceramic [17], or a bipolar hinge component and a stainless steel stem [16]. At present, because of its acceptable features, metal alloy—such as Ti alloy or cobalt–chromium (Co-Cr) alloy and a polyethylene liner—are common compositions in joint-prosthesis materials, especially for those in the lower limb. In my opinion, these materials are still the first choice for distal radial prosthesis at this time.

In theory, the aspects of stability and simulating biomechanical activities using distal radial prosthesis are poorer than total wrist-joint prosthesis, especially in the activity of forearm rotation. Moreover, more extensive soft tissue involvement would have a higher incidence of distal subluxation and dislocation. So, we sacrifice part of the activity of the prosthesis in order to maintain stability of the wrist joint. Further improvements in the prosthesis design should take full account of these problems, maybe like the prosthesis used by Natarajan et al. [16], which has the built-in joint for wrist mobility and can stabilize or reconstruct radiocarpal articulation. Late complications, such ad degenerative changes and aseptic loosening to the wrist joint following reconstruction with a distal radial prosthesis appear to be inevitable, as seen in studies with longer follow-up. Joint degeneration and loosening must also be taken into account in the upgrade in order to create a more appropriate prosthesis.

We recognize the following significant limitations of our results: the number of patients is small, the follow-up is not yet sufficient to report on long-term results, and it is retrospective. Larger series and a longer follow-up are needed to verify the long-term efficacy of this promising surgical technique. However, we believe that the desired outcome can be achieved with careful patient selection, precise preoperative workup, and meticulous surgical technique. In our opinion, the type of prosthetic replacement reported in this study is not suitable for patients with soft tissue infection, limited surrounding soft tissue, severe scarring, severe osteoporosis, or high functional demands of the hand.

Conclusion

Except for some limitations, in our opinion, this method results in a reasonable functional outcome at intermediate follow-up evaluation. Although average ROM of the wrist is poorer than reported in some studies using other techniques, custom prosthetic replacement can be considered a reasonable and alternative option after en bloc excision for Campanacci grade II/III GCT of the distal radius.

Compliance with ethical standards

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