



Management of elbow dysfunction associated with giant cell tumor of the distal humerus: achieving oncologic safety and good function by a combined reconstruction



Hao-Min Cui, MD^{a,b,1}, Ya-Ling Yu, MD^{a,1}, Yu He, MD^{c,1}, Ling-Zhi Kong, MD^a,
Cun-Yi Fan, MD, PhD^{a,b,*}

^aDepartment of Orthopedic Surgery, Shanghai Jiao Tong University Affiliated Sixth People's Hospital, Shanghai, China

^bDepartment of Orthopedics, Shanghai Sixth People's Hospital East Affiliated to Shanghai University of Medicine & Health Sciences, Shanghai, China

^cDepartment of Orthopaedics, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences & Peking Union Medical College, Beijing, China

Background: The purpose of our study was to evaluate the functional outcomes and oncologic results of elbow salvage surgery using arthrolysis combined with ligament repair and external fixation for reconstruction of the elbow after tumor excision and autografting.

Methods: We retrospectively reviewed 6 patients with elbow dysfunction associated with giant cell tumor of the distal humerus. All patients were treated with our combined protocol. We assessed the Musculoskeletal Tumor Society system score, range of motion, Mayo Elbow Performance Score, recurrence, and complications for each patient.

Results: The mean follow-up period was 48 months (range, 36-60 months). There were no cases of post-operative fracture, infection, elbow dislocation, elbow stiffness, or local recurrence. The average Musculoskeletal Tumor Society score was 28 of 30 points (93%; range, 87%-100%). The Mayo Elbow Performance Score improved from a mean of 61 points to 93 points, with mean flexion of 135° and mean extension of 3°.

Conclusions: Local tumor resection, autografting, and elbow reconstruction by arthrolysis combined with ligament repair and external fixation can be performed with oncologic safety and provide satisfactory functional outcomes with low complication rates.

Level of evidence: Level IV; Case Series; Treatment Study

© 2018 Journal of Shoulder and Elbow Surgery Board of Trustees. All rights reserved.

Keywords: Elbow dysfunction; giant cell tumor of bone; distal humerus; functional outcome; oncologic result; reconstruction; open arthrolysis

This study was approved by the Ethics Committee of Shanghai Sixth People's Hospital East Campus (No. DY-KY-2017-019).

*Reprint requests: Cun-Yi Fan, MD, PhD, Department of Orthopedics, Shanghai Jiao Tong University Affiliated Sixth People's Hospital, 600 Yi Shan Road, Shanghai 200233, China.

E-mail address: cyfan@sjtu.edu.cn (C.-Y. Fan).

¹These authors contributed equally to this work.

Giant cell tumor of bone (GCTB) is defined as a locally aggressive but rarely metastasizing tumor, accounting for approximately 20% of all benign tumors and 5%-6% of primary bone tumors.^{4,18} The reported morbidity rate in some Asian countries may be higher than that in Western countries.¹⁴ GCTB commonly occurs at the meta-epiphyseal region of long bones, especially at the distal femur, proximal tibia, and proximal humerus. It rarely arises in the distal humerus, with this location accounting for only 0.8% of total GCTB cases; the distal humerus location is likely to affect elbow function.^{9,14}

Patients with GCTB usually present with pain, limited range of motion (ROM), and swelling at the site of the lesion.⁷ The ideal aim of GCTB treatment is to excise tumor tissue, reconstruct the bone defect, and restore limb function. Surgery is the classic treatment. However, postoperative limb function is usually compromised owing to the periarticular location of GCTB,^{1,9} although nearly 100% local control can be accomplished by en bloc resection and adjuvant agents.^{17,19} Owing to the rarity of GCTB located in the distal humerus, the need to reconstruct the distal humerus or elbow for GCTB has not been investigated in many studies.

The anatomic uniqueness and functional requirements of the elbow make tumor excision and restoration of this region challenging. To achieve joint salvage and optimal functional outcomes, we used a combined protocol to address this clinical challenge in GCTB treatment. The aim of our treatment strategy was to excise the tumor by en bloc resection combined with restoration of elbow stability and activity. The purpose of this study was to report our experience with a surgical algorithm for GCTB of the distal humerus and evaluate the oncologic results, complications, and functional outcomes of the reconstruction method.

Materials and methods

Patients

Informed consent was obtained from all individual participants included in this retrospective study. We reviewed 6 patients with GCTB of the distal humerus treated surgically between 2004 and 2015. Details of the patients are shown in [Table I](#).

Preoperative imaging ([Fig. 1](#)) included radiographs of the lesion, computed tomography of the lesion, magnetic resonance imaging,

Table I Data and preoperative information of 6 patients

Patient No.	Sex/Age, yr	Diagnosis	Tumor localization	Campanacci grade	Neurologic symptoms	Preoperative extension/flexion, °	Preoperative supination/pronation, °	Preoperative MEPS
1	F/39	Prime GCTB	Distal medial humerus	II	No	30/110	60/90	60
2	M/51	Prime GCTB	Distal medial humerus	II	No	35/120	90/80	65
3	F/41	Prime GCTB	Distal medial humerus	II	No	20/105	90/90	60
4	M/24	Prime GCTB	Distal medial humerus	III	UN paralysis	40/120	90/80	50
5	F/44	Prime GCTB	Distal medial humerus	II	No	20/110	60/90	70
6	F/35	Prime GCTB	Distal medial humerus	II	No	30/120	70/90	60

MEPS, Mayo Elbow Performance Score; F, female; GCTB, giant cell tumor of bone; M, male; UN, ulnar nerve.

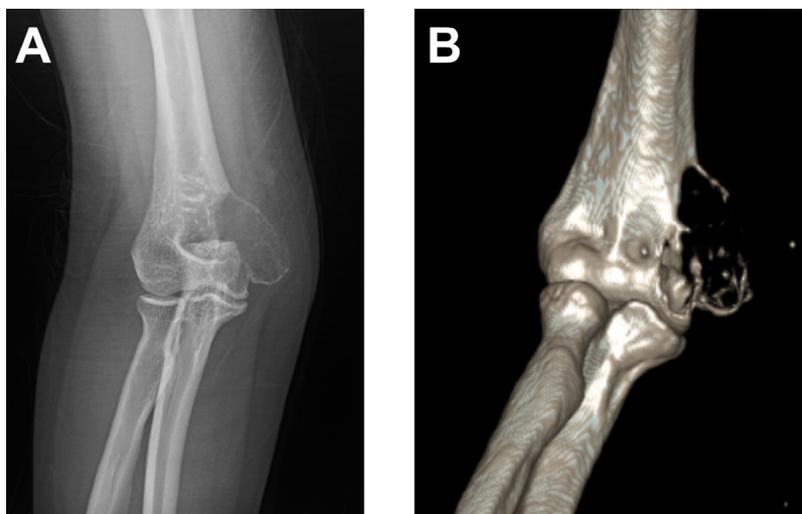


Figure 1 Imaging before index surgery. An anteroposterior radiograph (A) and 3-dimensional computed tomography scan (B) showed a giant cell tumor of the distal humerus.

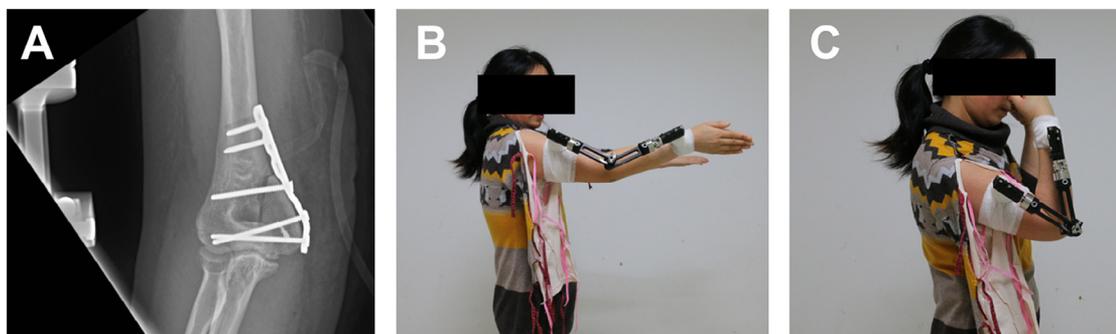


Figure 2 Application of combined reconstruction. (A) The postoperative anteroposterior view shows complete tumor resection and satisfactory elbow structure. (B, C) Extension and flexion with hinged external fixator in place.

nuclear scintigraphy, chest radiographs, and computed tomography of the chest. The giant cell tumor was graded according to the Campanacci staging system.³ Functional outcomes were analyzed using the Musculoskeletal Tumor Society (MSTS) system and Mayo Elbow Performance Score (MEPS).¹⁶ Elbow stability was assessed using the varus-valgus stress test and lateral pivot-shift test.

The pathologic diagnoses were obtained by needle or open biopsy. All lesions were histologically confirmed as benign GCTB. All patients' tumors were located at the medial condyle of the humerus, although this was not an inclusion criterion. This may result from the epidemiology of GCTB. No radiotherapy was performed before surgery. Mean preoperative extension was 29° (range, 20°-40°); the mean arc of flexion was 114° (range, 105°-120°). Mean preoperative supination was 77° (range, 60°-90°), and mean pronation was 87° (range, 80°-90°). All patients reported pain, and 1 patient had ulnar nerve paralysis with sensory deficit. Preoperatively, the mean MEPS was 61 points (Table I). All patients underwent a complex protocol including en bloc resection of the tumor site, autogenous iliac bone grafting, and arthrolysis, followed by fitting of a hinged external fixator.

Surgical procedure

Surgery was performed with the patient under general anesthesia with a sterile air tourniquet applied in the supine position. The operative technique primarily included excision (en bloc resection) of the tumor and reconstruction of elbow function. First, a medial approach was chosen, depending on the more affected side. After retaining a cuff of normal tissue and identifying and preserving the ulnar nerve, the surgeon exposed the elbow and completely removed the tumor. On the articular side of the humeroulnar joint, the subchondral bone was maintained. All tumors in this study were located at the distal medial humerus, and none involved the articular surface.

Second, at least 1 adjuvant, such as phenol, alcohol, or iodophor, was used in the procedures. Autogenous iliac bone graft was used to reconstruct the cavity after resection. After shaping, placement, and internal fixation of the iliac bone for reconstruction of the distal humeral defects, the arthrolysis procedure was performed.

Third, the posterior elbow was released through the medial approach. No further operation to release the ligaments was needed when the passive flexion-extension arc achieved ROM of 0° to 135° and the pronation-supination arc achieved ROM of 60° of pronation to 60° of supination. The Kocher approach was used to expose

and clean up the anterior capsule or coronoid fossa if necessary. The elbow ROM during the surgical procedure was recorded to guide postoperative exercises. In this case series, index surgery was performed via only a medial approach in 2 cases and via a combined lateral and medial approach in the others.

Fourth, owing to the en bloc resection of the tumor with soft-tissue extension, the medial collateral ligament was bluntly dissected from its origin. The normal ligament was reattached to the bone graft with the surrounding soft tissues by a bone suture anchor (Twifix; Smith & Nephew, Andover, MA, USA) (Fig. 2, A). Then, a unilateral hinged external fixator (Orthofix, Verona, Italy) was applied in all cases along the elbow rotational axis (Fig. 2, B and C), which was identified by C-arm radiography. Distraction of at least 3 mm was applied to each joint to protect the reconstructed ligament. In this case series, all patients underwent release of the ulnar nerve and subcutaneous transposition. The origins of the muscles were reattached, and the wound was closed in layers.

Postoperative treatment

Postoperatively, none of the patients required physiotherapy. No postoperative radiochemotherapy was applied in any patient. Celecoxib (200 mg once daily) was prescribed for 4 weeks to prevent heterotopic ossification (HO), and Methycobal (Eisai Co., Ltd., Tokyo, Japan) (0.5 mg 3 times per day) was prescribed if the patient had preoperative ulnar nerve paralysis. Active and passive postoperative movements were started at 24 to 48 hours postoperatively, and flexion-extension exercises were performed 3 times per day for 30 minutes each time during the first week postoperatively. After the first week, the duration of flexion-extension exercises increased to 60 minutes each time, and forearm rotation exercises were performed 3 times per day by temporarily removing the external fixator. Removal of the external fixator was performed at 6 to 8 weeks postoperatively as an outpatient procedure.

Statistical analysis

Statistical analyses of continuous variables, such as ROM and the MEPS, were performed using the independent-samples *t* test. Dichotomous variables were analyzed using the Fisher exact test. *P* < .05 was considered significant. Statistical analyses were performed with IBM SPSS Statistics for Windows (version 19.0; IBM, Armonk, NY, USA).

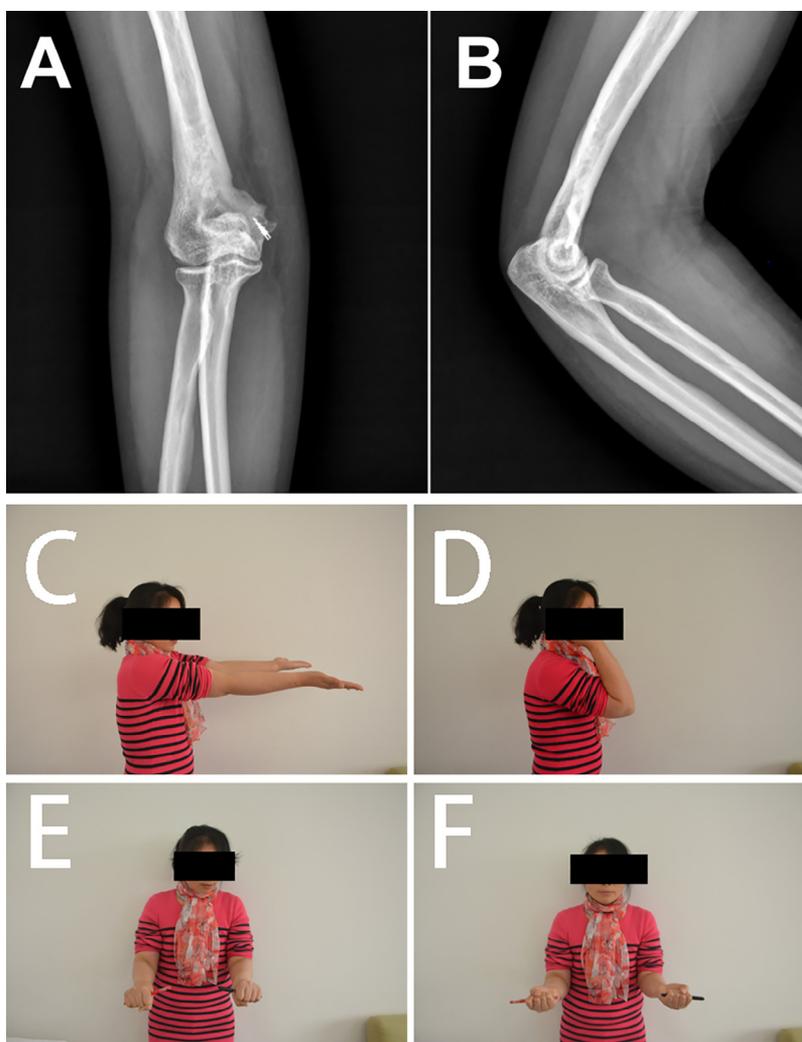


Figure 3 Elbow radiographs and function at 60-month follow-up. (A, B) The x-ray images show good anatomic structure and no sign of recurrence at last follow-up. Extension and flexion at external fixator removal (C, D) and at last follow-up (E, F).

Results

At most recent follow-up, all patients were alive and free from disease. The mean follow-up period was 48 months (range, 36-60 months). Local recurrence or distant metastasis did not develop in any patient. No operation was complicated by postoperative fracture, infection, elbow dislocation, or elbow stiffness. All bone grafting sites healed. No sign of extrusion of the screw or plate was radiographically detected during follow-up (Fig. 3, A and B).

At the final follow-up visit, all patients had an arc of elbow motion of more than 100° (Table II). Mean ROM increased from 85° preoperatively to 133° postoperatively, with mean flexion of 135° (range, 130°-145°; $P < .001$) and mean extension of 3° (range, 0°-10°; $P < .001$). Mean supination after the operation was 73° (range, 60°-90°; $P = .682$), and mean pronation was 85° (range, 70°-90°; $P = .313$). The MEPS improved from a mean of 61 points to 93 points ($P < .001$). The average MSTS score was 28 of 30 points (93%; range, 87%-100%) (Fig. 3, C-F).

Two patients complained of mild pain during activity. One patient who had preoperative ulnar nerve paralysis experienced persistent ulnar nerve symptoms (numbness of the little finger) postoperatively. A small degree of mature HO located extra-articularly was observed in 2 patients, but it did not affect joint function at the final follow-up. None of the patients required reoperation for surgical complications or tumor recurrence. Two patients underwent hardware removal of the plate according to their wishes at 28 months and 30 months postoperatively.

Discussion

Although GCTB is one of the most common primary musculoskeletal tumors, its ideal treatment has been a controversial and often discussed topic.^{1,4,13-15} The most important challenge in surgical management of GCTB is to provide optimal functional and oncologic results. Concerning tumors of the distal humerus, reconstruction of the functional elbow is technically difficult because it is an uncommon place for tumor

Table II Surgical information and outcomes of 6 patients

Patient No.	Surgical approach	Postoperative extension/flexion, °	Postoperative supination/pronation, °	Functional outcome		Follow-up, mo	Oncologic outcome	
				MEPS, points	MSTS score, %		Complications	Relapse
1	Medial	0/130	60/90	100	100	60	None	None
2	Medial and lateral	0/135	90/70	95	93	36	None	None
3	Medial	5/135	80/90	100	93	54	HO	None
4	Medial and lateral	0/145	80/80	85	87	48	UN paralysis	None
5	Medial and lateral	0/130	60/90	95	93	42	None	None
6	Medial and lateral	10/135	70/90	85	93	48	HO	None

MEPS, Mayo Elbow Performance Score; MSTS, Musculoskeletal Tumor Society; HO, heterotopic ossification; UN, ulnar nerve.

involvement. Most current studies of elbow reconstruction include various sites and different types of tumors, making it difficult to accurately compare oncologic and functional outcomes. Moreover, very few reports have focused on the management of elbow dysfunction associated with GCTB of the distal humerus.

It is necessary to develop an effective protocol in a targeted manner according to the special anatomic position and functional requirements of the elbow. Different scoring systems, including the MEPS, MSTS, and Toronto Extremity Salvage scoring systems, have been used in the functional analysis of surgical treatment of GCTB.^{10,14,16} In our study, both the MEPS and MSTS score were used to analyze the arc of motion and lifting ability, which are both essential factors for elbow functional evaluation. All patients underwent treatment with a protocol including complete eradication of the tumor, open release, and restoration of the elbow with bone graft combined with ligament repair and external fixation. The average MSTS score of 93% and MEPS of 93 points showed that this protocol could provide oncologic safety and good function with low complication rates. We attribute our good long-term outcomes to complete eradication of diseased tissue while preserving the normal bony architecture, as well as an aggressive protocol for restoration of stability and mobility of the elbow. The essential prerequisite for the limb salvage operation is radical eradication of tumor. Surgical treatment primarily includes curettage—sometimes extensive curettage—or resection.¹⁷ Previously published studies indicated that curettage was unable to significantly improve functional outcomes.^{2,11} Intralesional excision combined with curettage was previously considered the standard method of treatment but has been associated with high local recurrence rates ranging from 10% to 40%.^{2,11,12} En bloc resection or wide resection is well known to produce the lowest recurrence rate¹⁵; however, it carries a risk of poorer functional outcomes.⁸ Indications for en bloc resection include large tumor size with soft-tissue extension. Because type II or III GCTB is locally aggressive and destroys bone and overlying soft tissue, en bloc resection combined with local adjuvants was adopted in our protocol to prevent local recurrence. Our data showed that en bloc resection could produce a low relapse rate. However, the obtained results are not definitive because of the small sample size in our study.

The options for reconstruction after excision of a tumor in the elbow region are limited and disputed. Allografts were used to restore a range of distal humeral osseous defects caused by trauma and tumor in 23 patients reported by Dean and Urbaniak.⁶ Significant complications (69.5%), including deep infection (13%), elbow instability (26%), nonunion (21.7%), poor range of movement (8.7%), and graft resorption (4.3%), were reported in their study, and they concluded that “allograft was not recommended for routine use.” Hemiarthroplasty and arthrodesis are rarely undertaken and have an unpredictable outcome.^{10,16} Prosthetic replacement is another strategy for reconstruction that can improve elbow stability, but it has potential complications including the risk of prosthesis failure, which can be caused by aseptic loosening, infection, bushing failure, or periprosthetic fracture. In a case report describing 1 patient who underwent total elbow arthroplasty of the distal humerus for GCTB, the results showed ROM of 15°-120° and no evidence of recurrence at 18 months’ follow-up.⁹ Kulkarni et al¹⁰ described 10 patients with tumors of the distal humerus treated by endoprosthetic replacement. Four patients required additional surgery because of aseptic loosening and bushing failure. The mean fixed flexion deformity of the elbow was 15° and the mean flexion range was 115° after a mean follow-up of 8 years. Prolonged follow-up is necessary to determine whether elbow function, after replacement, deteriorates with time. For most of our cases, prosthetic replacement was considered too aggressive to be applied in patients with extra-articular tumors. Some reports used autograft for reconstruction of the bone defect and found no graft-related complications.¹⁵ Considering the ideal treatment goal of preservation of joint function and prevention of related complications, the bone defect after resection of GCTB was filled with shaped autogenous iliac bone graft and then strongly fixed with a supportive plate. During follow-up, all bone grafting sites healed and no graft-related complications occurred.

Restoration of joint function is another important objective of treatment. All elbows in our study showed ROM dysfunction of varying duration before surgery. After complete eradication of tumor, our treatment protocol including arthrolysis combined with ligament reconstruction and external fixation was introduced to rebuild stability and mobility of the elbow. Arthrolysis is often used for the treatment of

elbow post-traumatic stiffness^{5,20} but is less commonly reported in the treatment of tumors. Because of long-term dysfunction often resulting in contractures of the joint capsule or ligament and incomplete cleanup during elbow surgery resulting in HO, open arthrolysis is an indispensable part of our index protocol. Moreover, the application of arthrolysis after tumor resection resulted in good functional outcomes in our study.

GCTB often occurs in the metaphyseal region and invades surrounding tissues. In our study, after resection of the tumor and soft-tissue lesion, the remaining ligament lost its attachment point, resulting in an unstable elbow. Thus, the reconstruction of stability is vital to the postoperative recovery of elbow function. In this study, the normal surrounding soft tissue and ligament were reattached to the bone graft and reconstructed with a bone suture anchor. Then, an external fixator played an instrumental role in maintaining newly established stability and ensuring security during postoperative rehabilitation. At follow-up, joint stability was obtained in all cases, including 1 case presenting with mild valgus laxity (<10°). Evaluation of elbow stability showed that this combined strategy is a safe and effective means for the promotion of postoperative function.

The major limitations of our study are the small sample size, retrospective nature, and absence of a control group. In our study, all tumors were located in the distal medial humerus. This may be because of the predilection for GCTB of the distal humerus to be located medially. Thus, prospective studies with larger numbers of patients will be required to substantiate the findings of this study.

Conclusion

Combined use of local tumor resection, autografting, and reconstruction with elbow arthrolysis together with ligament repair and external fixation can provide oncologic safety and satisfactory functional outcomes.

Disclaimer

The authors, their immediate families, and any research foundations with which they are affiliated have not received any financial payments or other benefits from any commercial entity related to the subject of this article.

References

- Balke M, Schremper L, Gebert C, Ahrens H, Streitbueger A, Koehler G, et al. Giant cell tumor of bone: treatment and outcome of 214 cases. *J Cancer Res Clin Oncol* 2008;134:969-78. <http://dx.doi.org/10.1007/s00432-008-0370-x>
- Blackley HR, Wunder JS, Davis AM, White LM, Kandel R, Bell RS. Treatment of giant-cell tumors of long bones with curettage and bone-grafting. *J Bone Joint Surg Am* 1999;81:811-20.
- Campanacci M, Baldini N, Boriani S, Sudanese A. Giant-cell tumor of bone. *J Bone Joint Surg Am* 1987;69:106-14.
- Chawla S, Henshaw R, Seeger L, Choy E, Blay JY, Ferrari S, et al. Safety and efficacy of denosumab for adults and skeletally mature adolescents with giant cell tumour of bone: interim analysis of an open-label, parallel-group, phase 2 study. *Lancet Oncol* 2013;14:901-8. [http://dx.doi.org/10.1016/s1470-2045\(13\)70277-8](http://dx.doi.org/10.1016/s1470-2045(13)70277-8)
- Cui HM, Yu YL, He Y, Cheng Y, Liu JZ, Zheng W, et al. Management of elbow stiffness after postoperative treatment of terrible triad elbow injury: maintaining mobility and stability using a combined protocol. *Int Orthop* 2018;42:609-18. <http://dx.doi.org/10.1007/s00264-017-3721-x>
- Dean GS, Urbaniak JR. Elbow allograft for reconstruction of the elbow with massive bone loss: long term results. *Clin Orthop Relat Res* 1997;12-22.
- Gamberi G, Serra M, Ragazzini P, Magagnoli G, Pazzaglia L, Ponticelli F, et al. Identification of markers of possible prognostic value in 57 giant cell tumors of bone. *Oncol Rep* 2003;10:351-6. <http://dx.doi.org/10.3892/or.10.2.351>
- Gitelis S, Mallin BA, Piasecki P, Turner F. Intralesional excision compared with en bloc resection for giant-cell tumors of bone. *J Bone Joint Surg Am* 1993;75:1648-55.
- Hegde AS, Shenoy RM, Rai MP. Giant cell tumour of the distal humerus: a case report. *J Orthop Surg* 2014;22:427-9. <http://dx.doi.org/10.1177/230949901402200332>
- Kulkarni A, Fiorenza F, Grimer RJ, Carter SR, Tillman RM. The results of endoprosthetic replacement for tumours of the distal humerus. *J Bone Joint Surg Br* 2003;85:240-3. <http://dx.doi.org/10.1302/0301-620x.85b2.13524>
- Lausten GS, Jensen PK, Schjødt T, Lund B. Local recurrences in giant cell tumour of bone. *Int Orthop* 1996;20:172-6.
- Malek F, Krueger P, Hatmi ZN, Malayeri AA, Faezipour H, O'Donnell RJ. Local control of long bone giant cell tumour using curettage, burring and bone grafting without adjuvant therapy. *Int Orthop* 2006;30:495-8. <http://dx.doi.org/10.1007/s00264-006-0146-3>
- Mankin HJ, Hornicek FJ. Treatment of giant cell tumors with allograft transplants. *Clin Orthop Relat Res* 2005;439:144-50. <http://dx.doi.org/10.1097/01.blo.0000174684.85250.b5>
- Niu X, Zhang Q, Hao L, Ding Y, Li Y, Xu H, et al. Giant cell tumor of the extremity: retrospective analysis of 621 Chinese patients from one institution. *J Bone Joint Surg Am* 2012;94:461-7. <http://dx.doi.org/10.2106/JBJS.J.01922>
- Su YP, Chen WM, Chen TH. Giant-cell tumors of bone: an analysis of 87 cases. *Int Orthop* 2004;28:239-43. <http://dx.doi.org/10.1007/s00264-004-0564-z>
- Tang X, Guo W, Yang R, Tang S, Yang Y. Custom-made prosthesis replacement for reconstruction of elbow after tumor resection. *J Shoulder Elbow Surg* 2009;18:796-803. <http://dx.doi.org/10.1016/j.jse.2009.01.022>
- Van der Heijden L, Dijkstra PD, van de Sande MA, Kroep JR, Nout RA, van Rijswijk CS, et al. The clinical approach toward giant cell tumor of bone. *Oncologist* 2014;19:550-61. <http://dx.doi.org/10.1634/theoncologist.2013-0432>
- Werner M. Giant cell tumour of bone: morphological, biological and histogenetical aspects. *Int Orthop* 2006;30:484-9. <http://dx.doi.org/10.1007/s00264-006-0215-7>
- Yang Y, Huang Z, Niu X, Xu H, Li Y, Liu W. Clinical characteristics and risk factors analysis of lung metastasis of benign giant cell tumor of bone. *J Bone Oncol* 2017;7:23-8. <http://dx.doi.org/10.1016/j.jbo.2017.04.001>
- Yu SY, Wang W, Liu S, Ruan HJ, Liu JJ, Li XJ, et al. Arthrolysis and delayed internal fixation combined with hinged external fixation for elbow stiffness associated with malunion or nonunion of capitellum fracture. *J Shoulder Elbow Surg* 2015;24:941-6. <http://dx.doi.org/10.1016/j.jse.2015.01.019>