

Distal radioulnar joint prosthesis for the treatment of giant cell tumor of the distal ulna: a case report and literature review

Isidre Gracia · Ignacio R. Proubasta ·
Laura Trullols · Ana Peiró · Esther Moya ·
Sarah Cortés · Oscar Buezo · Joan Majó

Received: 10 August 2010 / Accepted: 9 July 2011 / Published online: 20 July 2011
© The Author(s) 2011. This article is published with open access at Springerlink.com

Abstract Giant cell tumor (GCT) of the distal end of the ulna is an uncommon site for primary bone tumors. When it occurs, en-bloc resection of the distal part of the ulna with or without reconstruction stabilization of the ulnar stump is the recommended treatment. We present a case of a 56-year-old man with a GCT of the distal ulna treated successfully with an en-bloc resection of the distal ulna with reconstruction using radioulnar joint prosthesis. Although the experience with this type of treatment is limited, implantation of a metallic prosthesis to replace the distal part of the ulna can also be considered as a salvage procedure for the treatment of this difficult pathology.

Keywords Distal ulna · Giant cell tumor · Resection · Radioulnar prosthesis

Introduction

Giant cell tumor (GCT) of bone is a rare, benign, but locally invasive neoplasm, accounting for 3–5% of all primary bone tumors [1, 2]. Usually, these tumors involve the distal femur, proximal tibia and distal radius of skeletally mature individuals [3]. However, the location of the distal ulna is very unusual, with a reported incidence from 0.45 to 6% [4–15]. When this occur, the treatment of choice is controversial. However, en-bloc resection of the distal part of the ulna with or without reconstruction stabilization of the ulnar stump is the recommended treatment

for distal ulna CGT [6–8], although in some cases with excessive resection of the distal ulna, residual pain due to impingement, impaired gripping and reduced load bearing are those most frequently experienced [15]. Is for this reason that the radioulnar joint prosthesis can be a valid option as a salvage procedure for the treatment of this difficult problem [16]. However, the clinical experience with this prosthesis after en-bloc resection of CGT in the distal ulna is limited, although it seems to be that the initial results are promising [10, 11, 13].

The purpose of this report is to review the literature of the distal ulna GCT and to present another case with this localization that was treated successfully with resection of a large distal ulnar segment followed by reconstruction with radioulnar prosthesis.

Case report

A 56-year-old man presented with a 6-month history of pain in his non-dominant left wrist. Although the range of motion of the wrist was normal, the distal end of the ulna was enlarged and tender. Radiographs revealed a multi-lobular and radiolucent area with a clear margin in the distal ulna (Fig. 1). A CT scan showed a lesion measuring 8 cm in length and 4 cm in width, expanding and partly destroying the thin cortex. On T1-weighted MRI images, the lesion showed a low-intensity signal and on T2-weighted images with fat suppression, a heterogeneous high-intensity signal (Fig. 2). Plain chest radiographs and a CT scan of the thorax and abdomen were normal without evidence of metastasis.

The work-up diagnosis was primary GCT, which was confirmed by trucut biopsy. The patient underwent a wide en-bloc resection of a GCT of the distal ulna, including the

I. Gracia · I. R. Proubasta (✉) · L. Trullols · A. Peiró ·
E. Moya · S. Cortés · O. Buezo · J. Majó
Orthopaedic Department, Hospital Santa Cruz Y San Pablo,
Barcelona, Spain
e-mail: iproubasta@santpau.cat

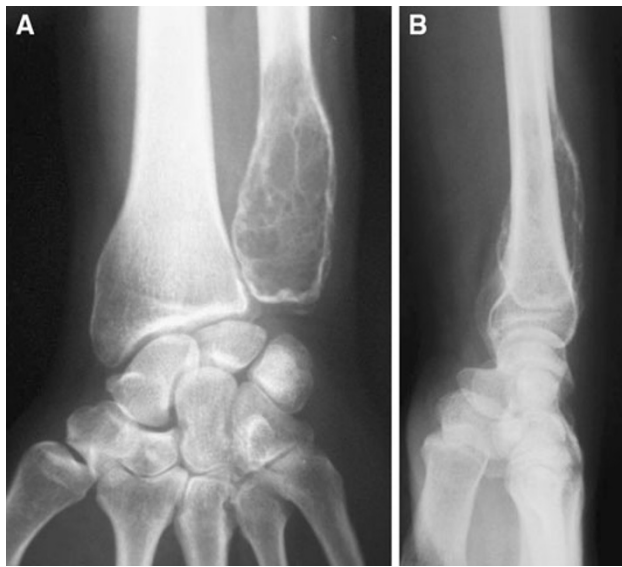


Fig. 1 Postero-anterior (a) and lateral (b) radiographs of the distal forearm and wrist show a multilobular and radiolucent area with a clear margin in the distal end of the ulna

triangular fibrocartilage complex, the ulnar border of the pronator quadratus and part of the distal radioulnar joint capsule, with immediate reconstruction of the distal radioulnar joint (DRUJ) using a radioulnar joint prosthesis (APTIS MEDICAL, Louisville, Kentucky, USA) [16]. This semi-constrained prosthesis was used to substitute the deficiency created by the absence of soft tissue support of the DRUJ and to address the gross instability of the remaining ulna caused by a large segmental resection. In our case, 2 cm of radiographically normal-appearing

proximal ulnar bone was resected in order to achieve a wide safety margin. The total amount of bone removed from the distal end of the ulna was 10 cm long. As a consequence of this, we placed one short diaphyseal fibular intercalary allograft between the ulnar stump and the ulnar stem prosthesis in order to cover the total surface of the plasma coating implant within the ulna (Fig. 3).

After the surgical intervention, we applied a well-padded short-arm splint. The splint remains in position for 2 weeks at which time sutures are removed and range of motion exercises are started. The tumor was sent for routine gross, histological and pathological examinations. Histologically, the lesion was a typical CGT composed of a large number of uniformly distributed osteoclast-like giant cells in a bland stroma of spindle-shaped mononuclear cells. Mitotic figures were absent, and surgical margins were free of tumor.

The functional results were evaluated using two scoring system: the Ferracini et al. score [7], based on range of motion, pain level, muscle strength and the presence or absence of ulnar impingement and ulnar or carpal instability and Mayo wrist score [17], based on pain intensity, functional status, range of motion and grip strength. With the Ferracini et al. [7] score, the patient achieved excellent results, scoring 18 out of 18 points at 6 and 12 months post-operatively, and with respect to the Mayo wrist score [17], the patient achieved a score of 82 (good) and 92 (excellent) at 6 and 12 months, respectively.

The oncologic results showed a good evolution with no evidence of local recurrence at 2-year follow-up. The patient's wrist was free from pain, and measurements of active range of motion, hand grip and key pinch strength at

Fig. 2 MRI of the wrist. On T1-weighted image with fat suppression, the lesion showed a low-intensity signal (a), while on T1 coronal turbo-spin-echo (TSE) weighted image (b), a heterogeneous high-intensity signal was observed

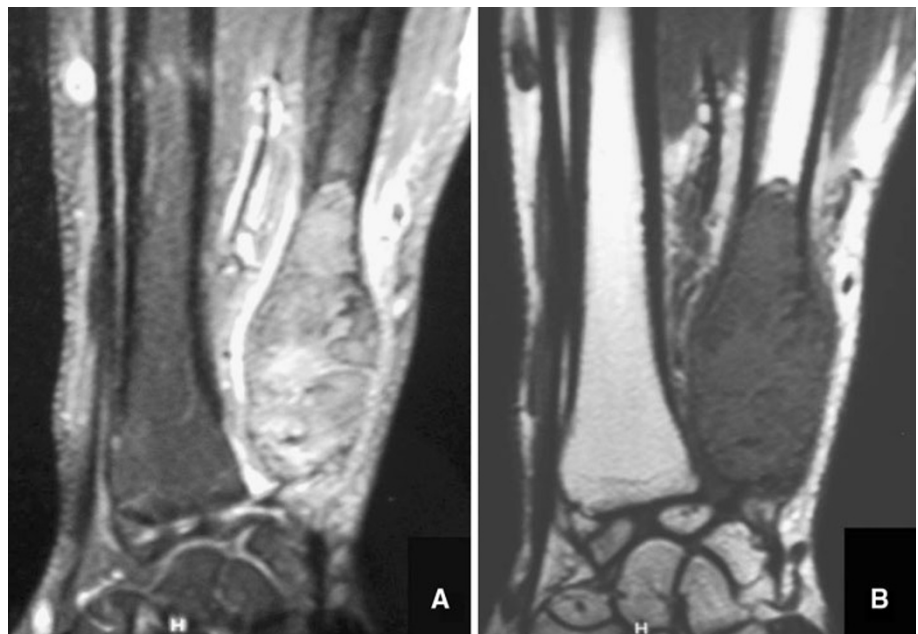




Fig. 3 Post-operative postero-anterior (a) and lateral (b) radiographs, showing the radioulnar joint prosthesis in place. Note the intercalary allograft between the ulnar stump and the ulnar stem prosthesis in order to stabilize the implant

Table 1 Range of motion of right and left wrist

Range of motion	Right	Left
Palmar flexion	80	80
Dorsiflexion	70	70
Ulnar deviation	40	35
Radial deviation	20	15
Supination	80	80
Pronation	85	80

Measurements were calculated in degrees using a goniometer 2 years post-operatively

2 years post-operatively are shown in Tables 1 and 2, respectively.

Discussion

Giant cell tumor of bone is rare. The reported annual incidence ranges from 0.65 to 1 cases per million population and affects people between 20 and 40 years of age and is more common in women than in men [18, 19]. Nearly 50% of the cases occur in the region of the knee. The distal radius is the third most common site for GCT, accounting

Table 2 Bilateral measurements of hand grip and key pinch strength (kg) were calculated using JAMAR dynamometer

	Right	Left
Hand grip strength	44.6	40.2
Key grip strength	14	13.4

Measurements are the average of three trials

for approximately 10% of cases [18, 20, 21]. However, the location of the distal ulna is very unusual, with a reported incidence from 0.45 to 6% [4–15]. When this occur, various treatment options have been proposed, including intralesional curettage, curettage and bone grafting, cryotherapy of the cavity after curettage, application of phenol after curettage, radiation, insertion of methylmethacrylate cement in the cavity after curettage, resection followed by allograft, en-bloc resection with or without reconstruction or stabilization of the ulna and prosthetic reconstruction [22]. Of these, en-bloc resection of the distal part of the ulna maintaining extra-lesional margins with or without reconstruction or stabilization of the ulnar stump is the more oncologically advantageous treatment for GCT located in this area [6–8]. According to Cooney et al. [6], reconstruction of the osseous defect after resection of a neoplasm of the distal end of the ulna, including GCT, is usually not necessary to maintain function. Similar results were observed by Wolfe et al. [8] in a multicenter study over wide excision of the distal ulna. However, other authors disagree because stabilization of the distal ulna following large resection, as a our case, can be a significant clinical problem with associated pain and weakness due to a decreased interosseous space with ulnar stump impingement on the radius metaphysis or instability of the radio-carpal joint with ulnar translation of the carpus [23–25]. For this reason, they suggest that soft tissue stabilization of the ulnar stump should be performed whenever possible. Although an oncological prosthesis per se is currently not available to reconstruct the distal ulna following the resection of a tumor, when all of the soft tissue support is removed from the distal ulna (fundamentally, the triangular fibrocartilage complex and the interosseous membrane), the radioulnar joint prosthesis can be an option in order to replace the gross instability of the remaining ulna caused by a large segmental resection [26]. In this sense, several prostheses have been designed for replacement of the DRUJ. However, most of them are designed to be used in patients with intact soft tissue and stabilizing ligaments at the DRUJ. These devices, therefore, are not appropriate for use in patients who have undergone resection of the DRUJ, because anteroposterior stability is not fully restored and suturing soft tissue limits mobility. However, the APTIS prosthesis can be used successfully to reconstruct and

Table 3 GCT of the distal ulna treated by en-bloc resection of distal ulna and reconstruction with radioulnar joint prosthesis. A literature review

Author	Year	Prosthesis	Follow-up	ROM						HG	KP	Result
				DF	PF	UD	RD	S	P			
Pirela-Cruz et al.	2008	APTIS [®]	13 m	70	65	45	24	70	80	40	12.8	E
Roidis et al.	2007	E-Centrix [®]	2 years			Full				ND	ND	E
Burke et al.	2009	STABILITY [®]	9 m			Full				ND	ND	E
Gracia et al.	2011	APTIS [®]	2 years	80	70	35	15	80	80	44.6	13	E

m months, *Y* year, *ND* not done, *DF* dorsiflexion, *PM* palmar flexion, *UD* ulnar deviation, *RD* radial deviation, *S* supination, *P* pronation, *E* excellent, *ROM* range of movement

stabilize the DRUJ following the resection of a large tumor, which in this case was a giant cell tumor of the distal ulna. Moreover, the fully constrained prosthesis provided the patient the ability to return to reasonable function following the resection of a large distal segment of the ulna, although other prosthesis models have been used with good results (Table 3).

Although conclusions cannot be drawn from some cases and long-term studies are required, radioulnar joint prosthesis for reconstruction after resection of a large segment of the distal ulna for GCT can be a valid option in order to reestablish the mechanical continuity of the forearm, reducing pain and improving strength and function.

Open Access This article is distributed under the terms of the Creative Commons Attribution License which permits any use, distribution and reproduction in any medium, provided the original author(s) and source are credited.

References

- McDonald DJ, Sim FH, Leod RA, Dahlin DC (1986) Giant cell tumour of bone. *J Bone Joint Surg* 68A:235–242
- Sung HW, Kuo DP, Shu WP, Chai YB, Liu CC, Li SM (1982) Giant-cell tumour of bone: analysis of two hundred and eight cases in chinese patients. *J Bone Joint Surg* 57A:167–173
- Dahlin DC, Cupps RE, Johnson EW Jr (1970) Giant cell tumor: a study of 195 cases. *Cancer* 25:1061–1070
- Blackley HR, Wunder JS, Davis AM, White LM, Kandel R, Bell RS (1997) Treatment of giant-cell tumours of long bones with curettage and bone grafting. *J Bone Joint Surg* 78A:811–820
- Goldenberg RR, Campbell CJ, Bonfiglio M (1970) Giant-cell tumour of bone. An analysis of two hundred and eighteen cases. *J Bone Joint Surg* 52A:619–664
- Cooney WP, Damson TA, Sim FH, Linscheid RL (1997) En bloc resection of tumours of the distal ulna. *J Bone Joint Surg* 79A:406–412
- Ferracini R, Masterson EL, Bell ES, Wunder JS (1988) Distal ulnar tumours. Results of management by en bloc resection in nine patients and review of the literature. *J Hand Surg* 23B:517–521
- Wolfe SW, Mih AD, Hotchkiss RN, Culp RW, Keinfhaber TR, Nagle DJ (1998) Wide excision of the distal ulna: a multicenter case study. *J Hand Surg* 23A:222–228
- Kayias EH, Drosos GI, Anagnostopoulou GA (2006) Resection of the distal ulna for tumors and stabilisation of the stump. A case report and literature review. *Acta Orthop Belg* 72:484–491
- Roidis NT, Gougoulias NE, Liakou PD, Malizos KN (2007) Distal ulnar implant arthroplasty as a definitive treatment of a recurrent giant-cell tumor. *J Hand Surg* 32A:1262–1266
- Pirela-Cruz MA, Higos M, Reddy K, Abdelfattah H, Cameron C, Hakim MN (2008) Treatment of a giant cell tumor of the distal ulna with a fully constrained prosthesis. *El Paso Phys Mag* 31:9–13
- Hashizume H, Kawai A, Nishida K, Sasaki K, Inoue H (1996) Ulnar buttress arthroplasty for reconstruction after resection of the distal ulna for giant cell tumour. *J Hand Surg* 21B:213–215
- Burke CS, Gupta A, Buecker P (2009) Distal ulna giant cell tumor resection with reconstruction using distal ulna prosthesis and brachioradialis wrap soft tissue stabilization. *Hand* 4:410–414
- Kawasoe Y, Yoshino S, Hayashi K, Yokouchi M, Onishi T, Komiyama S (2002) A case of giant cell tumor of the distal ulna. *Orthop Traumatol* 51:720–722
- James SLJ, Davies AM (2005) Giant-cell tumours of bone of the hand and wrist: a review of imaging findings and differential diagnoses. *Eur Radiol* 15:1855–1866
- Scheker LR, Babb BA, Killion PE (2001) Distal ulnar prosthetic replacement. *Orthop Clin North Am* 33:365–376
- Amadio PC, Berquist TH, Smith DK, Ilstrup DM, Cooney WP III, Linscheid RL (1989) Scaphoid malunion. *J Hand Surg* 14A:679–687
- Harness NG, Mankin HJ (2004) Giant-cell tumor of the distal forearm. *J Hand Surg* 20A:188–193
- Szendroi M (2004) Giant cell tumour of bone: a review article. *J Bone Joint Surg* 86B:5–12
- Campanacci M, Baldini N, Boriani S, Sudanese A (1987) Giant-cell tumour of bone. *J Bone Joint Surg* 69A:106–114
- Biscaglia R, Bacchini P, Bertoni F (2000) Giant cell tumours of the bones of the hand and foot. *Cancer* 88:2022–2032
- Blackley HR, Wunder JS, Davis AM, White LM, Kandel R (1999) Treatment of giant-cell tumors of long bones with curettage and bone-grafting. *J Bone Joint Surg* 81A:811–820
- Noble J, Arafa M (1983) Stabilisation of distal ulna after excessive Darrach's procedure. *The Hand* 15:70–72
- Laurentin-Pérez LA, Goodwin AN, Babb BA, Scheker LR (2008) A study of functional outcomes following implantation of a total distal radioulnar joint prosthesis. *J Hand Surg* 33E(1):18–28
- Bieber EJ, Linscheid RL, Dobyns JH, Beckenbaugh RD (1988) Failed distal ulna resection. *J Hand Surg* 13A:193–200
- Masaoka S, Longworth SH, Werner FW, Short WH, Green JK (2002) Biomechanical analysis of two ulnar head prostheses. *J Hand Surg* 27A:845–853