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CASE REPORT

Resection of the distal ulna for tumours and stabilisation of the stump. A case report and literature review

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The distal end of the ulna is an uncommon site for primary bone tumours. We report the case of a 23-year-old male, with a giant-cell tumour of the distal end of the ulna treated with en-bloc resection and stabilisation of the ulnar stump using one half of the extensor carpi ulnaris tendon. The amount of bone removed from the distal end of the ulna was 9.0 centimeters long. The functional and oncological results were excellent.

Stabilisation of the ulnar stump, using one half of the extensor carpi ulnaris tendon, has been described by Goldner and Hayes in 1979, after resection of a relatively small segment of the distal ulna. This is the first report on this technique for stabilisation of the ulnar stump after resection of a large distal ulnar segment. A literature review of reported cases with a resection of the distal ulna for primary bone tumours is presented. The available data are inconclusive as to whether a simple excision is adequate or a reconstruction/stabilisation is required.

Keywords : distal ulna ; giant-cell tumour ; resection ; stabilisation.

INTRODUCTION

The distal end of the ulna is an uncommon site for primary bone tumours. Giant cell tumour (GCT) of bone is a rare, benign, locally invasive tumour, accounting for approximately 3% to 5% of all primary bone tumours (*16, 23*). The location of GCT at the distal end of the ulna is very rare, with a reported incidence from 0.45% to 3.2% (*2, 10, 15, 21*). En bloc resection of the distal part of the ulna with or without reconstruction or stabilisation of the ulnar stump is the recommended treatment for GCT (*2, 4, 8, 9, 13, 14, 16*).

We present a patient with a GCT of the distal part of the ulna, treated with en-bloc resection of the distal ulna, followed by stabilisation of the remaining ulna using one half of the extensor carpi ulnaris (ECU) tendon. To our knowledge, this is the first report on the application of this technique after resection of a large distal ulnar segment.

The literature is reviewed and the various previously reported methods to treat distal ulnar tumours are discussed.

CASE REPORT

A 23-year-old, healthy appearing, right-handed male university student, presented with a 3-month history of painless, increasing swelling along the ulnar aspect of his left distal forearm. On examination the swelling was diffusely tender, and its

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Fig. 1. — Posterior-anterior radiographs of the distal forearm and wrist.

Fig. 2. — Lateral radiographs of the distal forearm and wrist

consistency was rather firm. The overlying skin was of normal colour and temperature. The range of motion of his left wrist was normal and painless. The grasping power was equal in both hands. There was no evidence of lymphadenopathy and no known systemic disease . Clinical examination did not reveal any abnormality. Haematological and biochemical investigations were normal.

Plain radiographs of the ulna showed an expanded multilocular and lytic lesion at its lower end, with at some places ill-defined borders, and no periosteal reaction (fig 1, 2). Bone scan showed a focally increased uptake in the distal ulna region, corresponding with the lesion on plain radiographs. CT scan showed a lesion measuring 5.4 cm in length and 3.4 cm in width, expanding and partly destroying the thin cortex (fig 3). On T1 weighted MRI images the lesion showed a low-intensity signal (fig 4) and on T2 weighted images with fat suppression, a heterogeneous high-intensity signal (fig 5). Abnormal tissue, demonstrating high-intensity signal on T2 weighted and uptake of the paramagnetic substance, possibly oedema, occupied the pronator quadratus muscle. Plain chest radiographs and a CT scan of the chest and abdomen were negative. The work-up diagnosis was primary GCT, which was confirmed by biopsy.

The extra-periosteal resection included the triangular fibrocartilage complex, the ulnar border of the pronator quadratus, although it was macro-

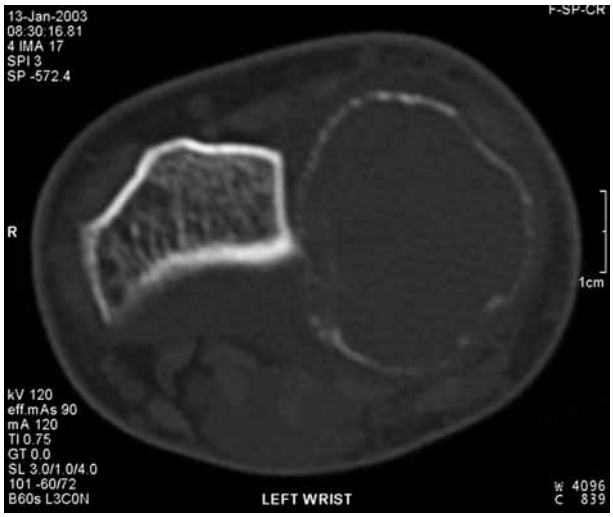

Fig. 3. — CT scan of the wrist

Fig. 4. — MRI of the wrist. T1 weighted image

Fig. 5. — MRI of the wrist. T2 weighted image

scopically free of disease and part of the distal radio-ulnar joint capsule. The sheath of the extensor carpi ulnaris (ECU) was easily dissected free from the mass by blunt dissection. The sheath was opened longitudinally, and then resected with the rest of the specimen, salvaging the ECU tendon. The remaining capsule was sutured and reinforced with the radial half of the ECU tendon, longitudinally split to a point 1 cm proximal of the new distal end of the ulna. The dorsal sensory branch of the ulnar nerve was preserved. The level of resection of the ulna was determined by the extent of the osseous involvement on radiographs and CT scan. An additional 3.5 centimeters of radiographically normal-appearing bone was resected in order to achieve a wide safety margin. The amount of bone removed from the distal end of the ulna was 9.0 centimeters long.

The decision was made to stabilise the ulnar stump, because of the age, the functional demands of the patient, and the high level of the resection. We chose the technique first described by Goldner and Hayes (*11*) in 1979 using one half of the ECU tendon, because there was a possibility that the tumour had extended anteriorly to the flexor compartment.

Fig. 6. — Posterior-anterior radiographs of the distal forearm and wrist at 3 years post surgery.

Fig. 7. — Lateral radiographs of the distal forearm and wrist at 3 years post surgery.

The tendon was passed through a 3-mm drill hole, 5 mm above the end of the ulnar stump in a dorsal to volar direction with the forearm held in supination. The tendon was then directed to the ulnar side and sutured back on itself. This created a cuff of tendon over the end of the ulnar stump, which stabilised the remaining ECU and tethered it toward the radial side of the ulna (fig 8). The wound was closed in a routine fashion over a drain.

Postoperatively the patient was immobilised in an open ulnar elbow splint with the forearm in supination for two weeks, following which physiotherapy commenced. The wrist was protected for three months with a removable splint at night and intermittent splinting during the day.

Histologically the lesion was a typical GCT composed of numerous osteoclast-like giant cells, interspersed with spindle-shaped stromal cells arranged in a storiform pattern. Mitotic figures were absent. Surgical margins were free of tumour.

The patient was seen every six months and at 3 years follow-up there was no evidence of recurrence or lung metastasis of the tumour. The functional result was evaluated using a simple scoring system described by Ferracini *et al* (*8*) based on range of motion, pain level, muscle strength, and the presence or absence of ulnar impingement and ulnar or carpal instability. The patient achieved excellent results, scoring 18 out of 18 points at 6 months postoperatively, with no instability of the stump or wrist.

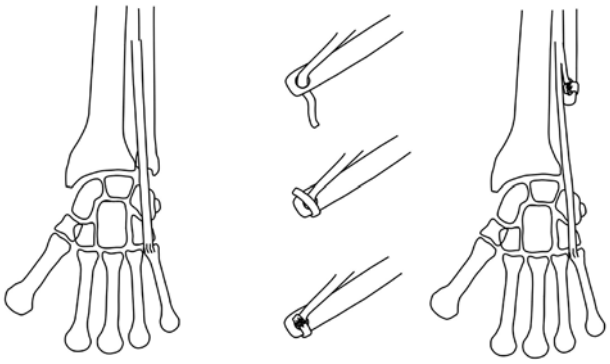

Fig. 8. — The tenodesis technique, first described by Goldner and Hayes (*11*) in 1979, using one half of the ECU tendon.

Changes in the angle between the long axes of the radius and third metacarpal shaft were not observed on radiographs at follow-up and thus showed no evidence of ulnar deviation of the carpus or ulnar subluxation of the carpus (fig 6, 7).

DISCUSSION

GCT of bone is rare. The reported annual incidence ranges from 0.65 to 1 case per million population (*12, 16*). Various treatment protocols involving wide or intra-lesional procedures have been developed for this often locally aggressive tumour.

Treatment options

Intralesional curettage without adjuvant therapy is associated with a high recurrence rate (*16, 19*). Several authors have used local adjuvants (eg, cytotoxic chemicals, cryotherapy, polymethylmethacrylate) in an attempt to obtain better control without resorting to more aggressive surgery such as wide resection.

Nevertheless, it seems more likely that adequate removal of the tumour rather than specific adjuvant treatment has more bearing on the recurrence rate (*27*).

En bloc resection of the distal part of the ulna, maintaining extra-lesional margins, with or without reconstruction or stabilisation of the ulnar stump, is the oncologically advantageous treatment for GCT located in the area (*14, 17*). En bloc (wide-intracompartmental) resection means removal of the entire tumour with its surrounding bone shell, periosteum, and reactive zone as a unit with a cuff of normal tissue (*6*) and involves excision of 25% to 50% of the ulnar length (6 to 11 cm) (*26*).

Distal ulna function

Functionally, the distal end of the ulna aids in rotation of the forearm, in grip strength as well as in maintaining the relationship between the carpus and the distal end of the radius. The ulnar collateral ligament of the wrist, which emanates from the ulnar styloid process and the triangular fibrocartilage complex, plays an important role in the maintenance of this anatomical relationship (*19, 20*).

Despite its functional importance, the distal ulna has always been considered to be dispendable. Its removal, with the classic Darrach procedure or its modifications (*5*) is performed for a variety of posttraumatic and degenerative conditions (*3*) with a documented failure rate of 10% to 50% (*5*).

Clinical experience with primary bone tumours of the distal part of the ulna is very limited, due to the low overall frequency. When faced with a benign locally aggressive neoplasm or a malignant tumour of the distal ulna, a wide resection is the recommended oncological procedure. A variety of additional treatment options has been reported, in an attempt to achieve a satisfactory functional result.

We reviewed the literature of osseous tumours of the distal ulna, to acquire a total view on the methods reported and the outcome of each method of treatment.

Simple resection

Some studies support the concept that wide resection of the distal ulna for an oncological reason, is a simple and durable treatment : the primary goal of excision of the tumour takes precedence over the restoration of function (*4*).

Cooney *et al* (*4*) advocated simple en bloc excision of the tumour in 9 cases of tumours of the

distal ulna. They concluded that reconstruction of the osseous defect with bone allograft, cement, a metallic prosthesis and even an autogenous bone graft adds unnecessary risk and morbidity without demonstrable functional gain.

Exner *et al* (*7*) reported two cases of malignant bone tumours (Ewing's sarcoma, osteosarcoma) presenting with similar clinical and comparable radiological and MRI features, treated with wide excision without reconstruction or stabilisation. Both patients were reported to have unimpaired hand function.

Wolfe *et al* (*26*) in a multicenter case study reported on 12 patients who underwent wide excision of the distal ulna without reconstruction for various conditions including 2 primary osseous neoplasms (parosteal osteosarcoma, desmoid tumour) with fair and excellent results respectively. They recommend wide excision of the distal ulna without reconstruction or stabilisation as the procedure of choice for distal ulnar neoplasms.

Harness and Mankin (*13*) reported on 3 patients with GCT of the distal ulna treated by primary resection. One of them underwent resection as a primary treatment and the remaining two as a salvage procedure after recurrence of the tumour following curettage and packing. All of the patients had a few complaints aside from some minor instability.

Reconstruction or stabilisation

On the other hand, many authors documented that wide excision of the distal ulna can be predicted to fail because of dorsal translation of the ulna at the resection site during pronation causing, (a) rupture of the digital extensor tendons (*18*), (b) pain and limitation of activity due to a decreased dynamic interosseous space with ulnar stump impingement on the radius metaphysis (*1*), (c) instability of the radio-carpal joint with ulnar translation of the carpus (*19*).

There are various factors that contribute to an increased likehood of unsatisfactory functional postoperative results in wide resections for tumours, compared to those obtained with the "standard" Darrach procedure such as : (a) the length of resection of the distal ulna for tumours is generally longer than for degenerative or post-traumatic conditions (*4*) and the rate of postoperative complications increases when resections are longer (*3, 18*), (b) the majority of resections for aggressive benign and malignant tumours of the distal ulna are extraperiosteal, thus precluding the stabilising role of the periosteal sleeve on the ulnar remnant, (c) the stabilising soft tissues surrounding the distal ulna are frequently removed as part of the excision of the tumour and this adds to the resultant instability and makes reconstruction more difficult. The adjacent soft tissues that may require excision include the wrist capsule, the triangular fibrocartilage complex (TFCC), the flexor carpi ulnaris (FCU), the extensor carpi ulnaris (ECU) tendons, the pronator quadratus and the interosseous membrane, and (d) tumour resections are performed in younger patients than the Darrach procedure, and the issue of stability is likely to be more important in this group (*1*).

Various methods for reconstruction or stabilisation have been reported after wide excisions of the distal ulna for primary osseous tumours including tendon stabilisation, distal ulnar autograft or allograft, plate arthrodesis and segmental bone transport using an Ilizarov fixator.

Ferracini *et al* (*8*) reported on 8 patients with tumours of the distal ulna, including 5 patients with GCT. They stabilised the ulnar stump with the flexor carpi ulnaris (FCU), fascia lata, with an autograft, or with plate arthrodesis. One case was treated without reconstruction and had a fair postoperative result. The authors concluded that soft tissue stabilisation of the ulnar stump should be performed whenever possible.

Gainor (*9*) reported on two cases of GCT and recommended a lasso tenodesis of the ulnar stump using a tendon graft from the palmaris longus (PL) to approximate the ECU and the FCU tendons to either side of the ulnar stump.

Hashizume *et al* (*13*) recommended an ulnar buttress arthroplasty to a housewife with a GCT of the distal ulna. The authors resected the distal ulna en bloc and grafted iliac bone to the ulnar side of the radius as a buttress using a screw and a K-wire. They reported excellent results at 6 months follow-up.

Wurapa and Whipple (*27*) reported on a patient with giant cell tumour of the distal ulna treated with wide en bloc excision and a 2-stage allograft reconstruction of the distal radioulnar joint. At 40 months follow-up the patient had good wrist function and was extremely satisfied with the procedure.

Stoffelen *et al* (*22*) reported on a patient with parosteal osteosarcoma of the distal ulna which was treated by wide resection and reconstruction by segmental transport of the remaining bone using an Ilizarov fixator. At the latest follow-up (3 years) the patient had an acceptable range of motion of wrist, elbow and shoulder but still an asymptomatic fibrous union of the distal ulna.

Goldner and Hayes (*11*), in 1979, originally described the extensor carpi ulnaris (ECU) tenodesis which we have used to stabilise the ulnar stump. They have operated on 225 patients for various conditions managed by resection of the distal ulna including post-traumatic and congenital deformities, rheumatoid arthritis as well as on a patient with osteochondroma. They also used this procedure to stabilise the ulnar stump after resection of the distal ulna for an osteochondroma, but no further details regarding the length of the resection are mentioned.

Our case is the first report of using this technique for stabilisation of the ulnar stump after resection of a large distal ulnar segment.

CONCLUSION

A literature review of reported cases with a resection of the distal ulna for primary bone tumours, is inconclusive as to (a) whether stabilisation or reconstruction is required or not and (b) as to the optimal method of stabilisation or reconstruction, if chosen. This is because prospective controlled trials are lacking comparing in a large number of patients, the efficacy of the various methods of primary excision of the distal ulna with or without stabilisation for tumours,

We believe that the optimal solution at this moment is stabilisation of the ulnar stump after en bloc resection of the distal ulna for a primary osseous tumour and its technique should be individualised, according to the age of the patient, the location and extension of the tumour, and the functional demands, without adding unnecessary risk and morbidity. An established diagnosis and careful treatment planning according to the oncologic standards are absolutely necessary before one proceeds to resection of what is thought to be a dispendable segment of bone with a benign or malignant tumour.

Although conclusions cannot be drawn from one case, stabilisation of the remaining ulna using one half of the ECU tendon, after excision of a large part of the distal ulna for a GCT, gave an excellent result.

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