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Case Report

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Reconstruction of Distal Radioulnar Joint after Resection of Giant Cell Tumour of Distal Ulna

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Abstract

Giant cell tumour is a rare, generally benign and locally aggressive tumour. It represents approximately 3% to 5% of all primary bone tumours. It usually occurs in skeletally mature individuals, between the ages of 20 and 40 years. Giant cell tumour of the distal ulna is extremely rare, accounting for approximately 0.45 to 3.2% of all the cases. As most of these tumours are locally aggressive, wide resection of the distal ulna with maximum preservation of adjacent joint function is the recommended treatment for giant cell tumour in such locations. Although some authors suggests only wide resection without reconstruction or stabilization of the ulnar stump that can cause wrist and distal radioulnar joint instability leading to pain, weakness, and loss of grip strength. To overcome those limitations, various reconstructive procedures have evolved. We think that reconstruction of distal radioulnar joint is of great importance, especially in young active patients. That's way we present a case of 21-year old patient with GCT of distal ulna treated with wide resection and reconstruction of the distal radioulnar joint with autologous free fibular graft and autologous palmaris longus tendon graft.

Keywords: *Giant Cell Tumour; Ulna; Resection; Reconstruction; Stability; Distal Radioulnar Joint*

Abbreviations: *GCT: Giant Cell Tumour; DRUJ: Distal Radioulnar Joint; TFCC: Triangular Fibrocartilage Complex*

Introduction

Giant cell tumour (GCT) is a rare, generally benign and locally aggressive tumour. It represents approximately 3% to 5% of all primary bone tumours [1]. It usually occurs in skeletally mature individuals, between the ages of 20 and 40 years and a female/male ratio of 1.2:1 [1,2]. It occurs in the ends of long bones in 75–90%. Most common sites of involvement are the distal femur (30%), proximal tibia (25%), distal radius (10%), and proximal humerus (6%) [3,4]. GCT of the distal ulna is extremely rare, accounting for approximately 0.45 to 3.2% of all the cases [3,5-8]. As most of these tumours are locally aggressive, wide resection of the distal ulna with maximum preservation of adjacent joint function is the recommended treatment for GCTs in such locations [8]. Although some authors suggests only wide resection without reconstruction or stabilisation of the ulnar stump others think that loss of ulnar support results in wrist and distal radioulnar joint (DRUJ) instability leading to pain, weakness, and loss of grip strength [9-11]. To overcome these limitations, various reconstructive procedures have evolved. We present a case of our 21-year old patient with GCT of distal ulna treated with wide resection and reconstruction of the distal radioulnar joint with autologous free fibular graft and palmaris longus tendon graft.

Case Report

A 21-year old male student presented in our Department with pain in the distal part of his right forearm after fall on the stretched arm during handball game. X-ray images showed expansive formation in the distal part of the right ulna (Figure 1). Detailed history revealed that he had pain and swelling in the distal, ulnar part of his forearm even 3 months earlier. Computed tomography, magnetic resonance imaging and bone scan, followed by biopsy, were performed in our Department. Computed tomography showed an expansive formation of the ulna, which was partially septated and affected approximately 7 cm of the distal ulna (Figure 2).

Figure 1: Radiogram (AP) of right wrist and distal forearm showing expansive tumour of distal ulna

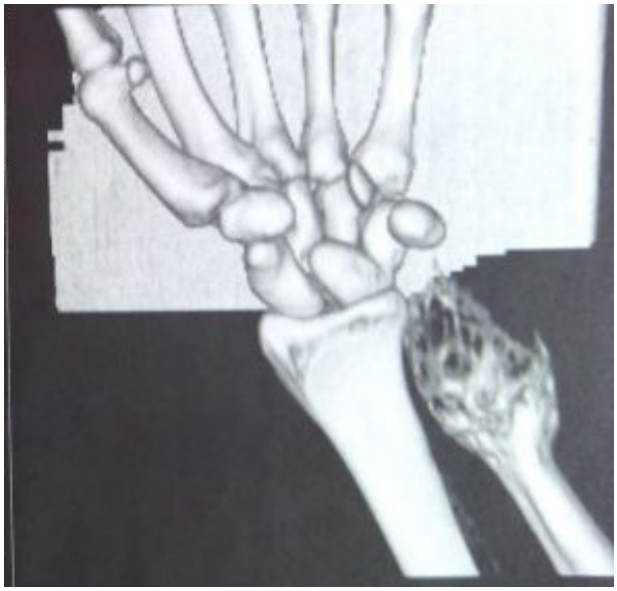

Figure 2: Computed tomography (3D reconstruction) showing expansive tumour of distal ulna

The tumour elevated periosteum of the ulna and broke through the cortex. The contour of the pisiform and the triquetrum were unclear with loss of cortical bone. Magnetic resonance imaging confirmed the presence of tumour expanding to the subcutaneous fat pad which wasn't involved. Bone scan showed increased pathologic perfusion of the distal ulna. Pathohistological findings showed very invasive growth of the tumour with lots of mitosis and penetration of the tumour trough whole thickness of cortical bone. Due to very aggressive, infiltrative and destructive growth of the tumour, especially in light with radiologic correlation we decided for two stage procedure. First we performed wide “en bloc” resection of distal 9 cm of ulna through standard approach to distal ulna between tendons of extensor and flexor carpi ulnaris including biopsy tract and skin scar from first operation. Complete triangular fibrocartilage complex (TFCC) was excised, as well as triquetrum and pisiform because imaging methods showed their destruction. Pisiform was stripped of flexor ulnaris tendon and the continuity of the tendon was preserved through the fibrous reflexion. Cement was moulded to resemble normal ulna and placed in to the forearm to maintain space for later reconstruction (Figures 3 and 4).

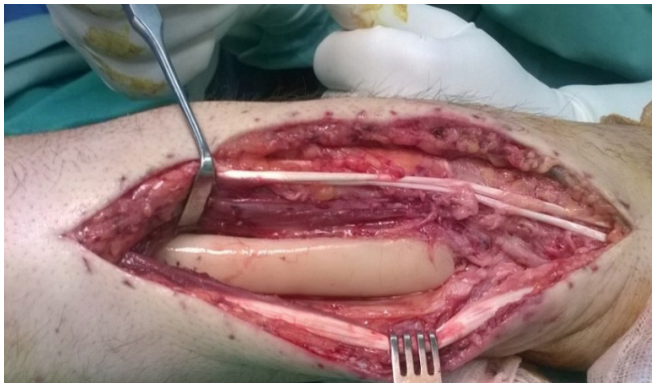

Figure 3: Cement spacer for temporary reconstruction of bone defect in first stage surgery

Figure 4: Radiogram after resection and implanting cement spacer

Wrist and forearm were immobilized in cast. Definitive pathohistological finding was GCT grade I-II, with lots of mitosis, but no pathological mitosis. There were vascular invasion of tumour and penetration of cortical bone, showing very aggressive nature of tumour. Proximal 2.5 cm of resected ulna and intramedullary canal were free of the tumour. Triquetrum and pisiform showed no signs of tumour. Six weeks after resection, second stage reconstruction procedure was performed. First, non-vascularised fibular shaft graft from patient right leg in length of 9 cm was prepared in standard manner from middle third of the fibula. Then we harvested palmaris longus tendon graft through two separate incisions, one at the level of wrist and the other 12 cm proximally on the forearm volar surface. Through the scar from previous operation we removed cement spacer. Then we matched the fibular graft and oriented it to best fit the ulnar stump and determined the place for the 3.5 mm DC plate. As fibular bone is very firm, because it is mainly consisted of cortical bone in the middle third, we performed the drilling and plating of the fibular graft on back table. Then we placed the graft with the plate and prepared eccentric holes in distal part on ulnar stump for compression and finished osteosynthesis (Figure 5). DRUJ was reconstructed with palmaris longus tendon with modified Adams procedure [12] (Figure 6). Skin incision was enlarged in dorsal, radial direction in distal part to facilitate exposure of distal radius. A drill holes were made in distal radius from volar to dorsal, located 5 mm proximally to the radiocarpal and 5 mm radially to sigmoid notch. It was prepared before fixing the fibular graft, because in that manner it was possible to easily reach palmar aspect of the radius. After fixing the fibular graft, palmaris longus tendon was passed through the hole in the radius, and then both ends were passed through the hole in the fibula and around the graft and then sutured to itself. In order to compensate triquetrum and pisiform bone loss and stabilize the ulnar side of the wrist, interposition arthroplasty using half of the extensor carpi ulnaris tendon was done. Wrist and forearm were immobilized in Sarmiento forearm cast in neutral position of the forearm which allowed flexion and extension in the elbow but blocked pronation and supination.

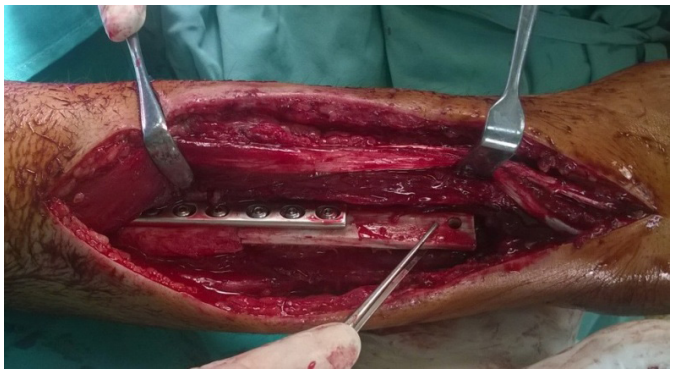

Figure 5: Distal ulna reconstruction with autologus free fibular graft and fixation with DC plate and screws

Figure 6: Stabilisation of DRUJ with palmaris longus tendon graft

Three years postoperatively, patient is feeling well, he has no pain, and he has satisfactory wrist range of motion, full pronation and supination of the forearm, full dorsal flexion of the wrist, palmar flexion 60 degrees and, ulnar deviation 45 and radial deviation 25 degrees (Figure 7). His grip strength is very similar to the contralateral side. In meantime he graduated and has no major problems with his hand in everyday activities. Control X-rays shows adequate integration of fibular graft at the site of osteosynthesis, with no signs of tumour recurrence (Figure 8). DRUJ is stable and pain free. DASH score during three years follow up improved to 12.5, compared to 54.2 preoperatively.

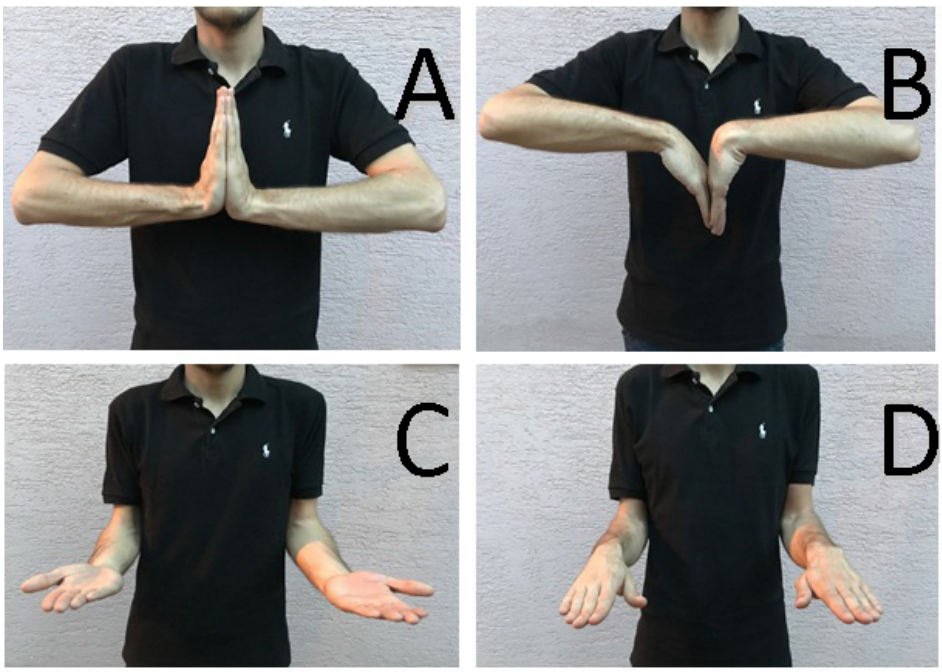

Figure 7: Range of motion three years after operation

Figure 8: Postoperative radiogram of right wrist and forearm

Discussion

The distal ulna and DRUJ play an important role in function of the wrist and the forearm. In relationship with the carpal bones and the distal end of the radius they transfer loads. Proximal and distal radioulnar joints allows pronation and supination of the forearm. The soft tissue structure, the triangular fibrocartilaginous complex (TFCC) is in charge of stability of DRUJ and grip strength.

GCT of the distal ulna has aggressive nature with high rate of recurrence. Before, many surgeons tried to retain the ulna and perform only curettage and packing cancellous bone with polymethylmethacrylate [6,8,13,14]. Unfortunately, there is a high recurrence rate of up to 40% when treated in such a manner [8]. Today recommended treatment option in those cases is wide "en bloc" resection [15]. Some authors have reported good functional outcome after only wide resection of the distal ulna without any kind of reconstructive procedure [2,16,17]. Others suggest only tenodesis of the ulnar stump with or without distal radioulnar stabilization by the modified Sauve-Kapandji procedure [18-20]. Although Darrach reported that the distal ulna can be excised without any functional limitations this procedure should be reserved for degenerative conditions, because failure rate of that procedure has been documented to be 10–50% [21,22]. Other authors revealed problems of loss of the ulnar support of carpal bones and ulnar stump instability after wide resection of the distal ulna [8,18,19,23]. That is especially important in young people with high functional demands. In order to preserve stability some authors use distal radioulnar prosthesis with satisfactory functional results [13,24,25]. Others prefer biologic reconstruction with bone graft and same kind of DRUJ stabilization [25]. We also think that both reconstruction of the DRUJ and stabilization of the ulna are equally important for functional rehabilitation after wide resection of the distal ulna.

In our young patient we decided for distal ulna reconstruction with autologous free fibular graft, and what is even more important stabilization of DRUJ to allow him the best possible range of motion and wrist function. We made interposition arthroplasty using half of the extensor carpi ulnaris tendon for removed triquetral bone. In that way we made spacer for lost carpal bone which preserved tension of remaining ligaments and capsule, and a "cushion" between carpus and free bone graft. We didn't find this procedure in literature but it was important to get as much stability and function so that seemed to be logical solution.

As far as we know only one similar case was published in the literature. Mariappan, et al., used proximal part of fibula as free graft for reconstruction of DRUJ and it was stabilised with palmaris longus graft with the sling of the extensor carpi ulnaris [25]. Although proximal part of fibular graft can potentially be better regarding cartilage cover of new joint surface, we don't feel comfortable destroying other important joint in the body.

Although we can't make conclusions based on one case, functional result in our patient shows us that reconstruction of DRUJ is of great importance after "en bloc" resection of GCT of distal ulna, especially in young active patients.

Conclusion

Giant cell tumour of the distal ulna is rare entity therefore there are no clear cut guidelines about the best way of treatment. Most of the authors agree that wide "en bloc" resection of distal ulna should be done. Recommendations about reconstructions are different, but we think that biologic reconstruction with obligatory DRUJ stabilisation could be the method of choice for young active patients.

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