

CASE STUDIES STUDII DE CAZ

Rehabilitation after total shoulder arthroplasty for a giant-cell tumor of bone - a case report

Reabilitarea după artroplastia totală de umăr pentru tumoră cu celule gigante a osului - prezentare de caz

Alina Popa¹, Alexandrina Nicu¹, Monica Borda^{1,2}, László Irsay^{1,2}, Rodica Ungur^{1,2},
Ioan Onac^{1,2}, Viorela Ciortea^{1,2}

¹Clinical Rehabilitation Hospital, Cluj-Napoca, Romania

²Rehabilitation Department, "Iuliu Hațieganu" University of Medicine and Pharmacy, Cluj-Napoca, Romania

Abstract

Background. Total shoulder arthroplasty is a standard operative treatment for a variety of disorders of the glenohumeral joint. Successful outcome of total shoulder arthroplasty depends on a well-designed and a well-executed physical therapy program. The rehabilitation program needs to respect a logical sequence: tissue healing, joint mobility and muscle strength.

Aims. The purpose of this study was to emphasize the best therapeutic options to functionally rehabilitate a shoulder that was surgically replaced after a giant-cell tumor excision.

Methods. The case report describes a 24-year-old female patient who presented in September 2013 to the Clinical Rehabilitation Hospital in Cluj-Napoca for rehabilitation after undergoing total shoulder arthroplasty for a giant-cell bone tumor located in the proximal region of the right humerus. Although they are considered benign in 90% of the cases, giant-cell tumors show a tendency for significant bone destruction, local recurrence, and occasional metastasis in the lung or lymph nodes.

The objective examination of the patient at her first hospital admission: normal weight (BMI=19 kg/m²), two postoperative scars in the anterior region of the right arm with minimal sensitivity when palpated, hypotonia and hypotrophy of the right upper limb, spontaneous pain at the mobilization of the right shoulder and limited active and passive range of motion (especially for flexion, abduction and external rotation), pain on the VAS = 30 mm, Constant Shoulder Score = 42, DASH score = 35, SPADI score = 53%. In our hospital, she underwent the current Neer protocol for postoperative total shoulder arthroplasty rehabilitation, which is widely used and is based on the basic science of soft tissue and bone healing.

Results. The functional outcome was good, as the range of motion and strength of the upper right limb improved after attending our physical therapy program.

Conclusions. When a well-performed surgical procedure is supplemented with a well-designed and frequently monitored therapy program, an excellent outcome of shoulder replacement should be expected.

Key words: rehabilitation, shoulder arthroplasty, giant-cell tumor.

Rezumat

Premize. Artroplastia totală de umăr reprezintă o intervenție chirurgicală standard pentru diverse afecțiuni ale articulației gleno-humeroale. Succesul endoprotezării de umăr depinde de calitatea programului de reabilitare instituit cât mai precoce. Programul de reabilitare trebuie să respecte o secvență logică: vindecarea țesuturilor, mobilitatea articulației și forța musculară.

Obiective. Obiectivul acestui studiu este de a pune în evidență cea mai bună conduită terapeutică pentru reabilitarea funcțională a unui umăr endoprotezat, după excizia unei tumori cu celule gigante.

Metode. Prezentarea de caz aduce în discuție cazul unei paciente în vârstă de 24 ani, care s-a prezentat în septembrie 2013 în Spitalul Clinic de Recuperare Cluj Napoca pentru recuperarea mobilității umărului drept după implantarea unei endoproteze totale la acest nivel pentru o tumoră cu celule gigante a osului, localizată în regiunea proximală a humerusului drept. Chiar dacă sunt considerate benigne în 90% din cazuri, tumorile cu celule gigante tind să evolueze spre distrucție osoasă, recidivă locală și, ocazional, metastaze în plămân și ganglionii limfatici.

La examenul obiectiv la prima internare se evidențiază: pacientă normoponderală (IMC=19 kg/m²), două cicatrici postoperatorii în curs de vindecare la nivelul feței anterioare a brațului drept, cu minimă sensibilitate dureroasă la palpare, hipotonie și

Received: 2015, February 17; Accepted for publication: 2015, February 20;

Address for correspondence: "Iuliu Hațieganu" University of Medicine and Pharmacy Cluj-Napoca, Clinical Rehabilitation Hospital, Rehabilitation Department, No. 46-50, Viilor St. 400437 Cluj-Napoca

E-mail: viorela.ciortea@yahoo.com; alina_med87@yahoo.com; monicampop@yahoo.fr

Corresponding author: Ileana Monica Borda, monicampop@yahoo.fr

hipotrofie a membrului superior drept, limitarea mobilității umărului operat (mai ales pentru flexie, abducție și rotație externă), durerea pe scala VAS = 30mm, Scorul Constant = 42, scorul DASH = 35, scorul SPADI = 53%. În spitalul nostru, pacienta a urmat protocolul curent Neer pentru reabilitarea umărului după artroplastie, care se bazează pe vindecarea țesuturilor moi și a osului.

Rezultate. Evoluția după programul de reabilitare a fost favorabilă, cu creșterea amplitudinii de mișcare și a forței musculare la nivelul membrului superior drept.

Concluzie. Când o intervenție chirurgicală de implantare a unei endoproteze este continuată de un program de reabilitare instituit precoce, bine controlat și individualizat, prognosticul funcțional este favorabil.

Cuvinte cheie: reabilitare, artroplastie de umăr, tumoră cu celule gigante.

Introduction

Total shoulder arthroplasty (TSA) is a standard operative treatment for a variety of disorders of the glenohumeral joint (Boudreau et al., 2007). Complications after TSA may include infection, instability, neurovascular injury, stiffness, cuff tear, periprosthetic fractures, glenoid erosion and component loosening (Sanchez-Sotelo et al., 2011).

The success of TSA depends on surgery, i.e. on the correct placement of the prosthesis, as well as on the quality of the rehabilitation program initiated as early as possible, which needs to respect a logical sequence: tissue healing, joint mobility and periarticular muscle strength (Brems, 2007).

It is also critical for the surgical team to communicate to the rehabilitation team important and relevant information about the surgical procedure, to ensure proper progression, so that the specific exercises are not advanced too rapidly in efforts to quickly provide functional rehabilitation (Cahill et al., 2014).

Rehabilitation after TSA is more challenging when the integrity of the rotator cuff is poor and the functional outcome is generally not as good as it is for patients with an intact rotator cuff (Frankle et al., 2005). Hawkins et al. (1989) concluded based on a series of 65 patients, followed up over an average of 40 months, that the underlying etiology of the disease process and the status of the rotator cuff are the best predictors of outcome for individuals treated with TSA.

Giant-cell tumors (GCT) are osteolytic primitive tumors with a rich vascularization, consisting of two predominant cell types: many multinuclear osteoclastic giant cells and mononuclear stromal cells, whose cytological appearance reflects the degree of aggressiveness, establishing in this way histological prognosis (Formasier et al., 1996). GCT represent 20% of all benign tumors and about 5% of primary bone tumors (Niu et al., 2012). The tumor commonly occurs during the second to fourth decades of life, with a female-to-male ratio of 1.3-1.5:1 (Schajowicz et al., 2001), and it usually originates from the epiphysis of long bones (Szendrői, 2004).

The radiographic appearance of the GCT is that of a lytic lesion with a well-defined non-sclerotic margin, eccentric in location, which extends near the articular surface. However, GCT may present aggressive features, such as cortical expansion or destruction of the soft-tissue components (Chakarun et al., 2013) and has a potential risk of local recurrence (0-65%) depending on the type of treatment (Klenke et al., 2011). Magnetic

resonance imaging (MRI) is often performed to evaluate the extent of the tumor. In typical GCT, the signal intensity is homogeneous, and the lesion is well circumscribed, with low signal intensity on T1-weighted images and intermediate signal intensity on T2-weighted images.

Being aggressive and potentially malignant lesions, GCT pose a challenging problem in accurately predicting their tendency to recur or metastasize. It is considered that 80% of GCT have a benign course, with a local recurrence rate of 10-50%, and about 10% undergo malignant transformation through their recurrences. The principal aim of the management of GCT is to eliminate the tumor and still save the joint function (Rahim et al., 2015). It is advised that each patient with GCT should be treated individually. Regardless of non-malignant features, the local behavior of tumors determines the treatment approach according to treatment principles for malignant tumors of bone (Maric et al., 2012).

Surgical and oncological treatment takes into account tumor location, previous treatments (in case of recurrence), histological aggressiveness and the presence or not of lung metastases, which are a cause of death in 16-25% of the cases (Kay et al., 1994). Surgical treatment consists of curettage and filling with cancellous bone or acrylic cement or mixed filling, in inactive forms (Kivioja et al., 2008). In case of recurrence, if histological examination evidences an inactive appearance, curettage is repeated, and if an active or aggressive appearance is evidenced, tumor resection will be performed (Becker et al., 2008). In case of an aggressive tumor, oncologic resection with or without reconstruction, depending on the tumor site (bone reconstruction or modular prosthesis), is carried out. Amputations or disarticulations are indicated in untreatable cases (Klenke et al., 2011).

The postoperative rehabilitation program for patients with TSA has a long duration (12-18 months), is intense, sustained and regular in nature, and includes the following main objectives: increase of the quality-of-life index, maximization of mobility in the replaced shoulder, as well as restoration of motor control in the entire upper limb. It is based on the protocol initiated by Neer, structured in three phases.

Phase I of the Neer protocol has the following main objectives: wound healing, maintenance of the integrity of the replaced joint, gradual increase in the passive range of motion of the shoulder, initiation of active elbow and wrist movements, reduction of pain and inflammation, reduction of muscle inhibition. The methods used are continuous orthosis use for up to 4 weeks, avoidance of shoulder overextension in clinostatism, avoidance of active shoulder

movements, weight lifting and rotations, cryotherapy, passive and self-passive shoulder mobilization, passive, self-passive, isometric and balancing exercises (Coodman type).

The objectives of phase II of the Neer protocol are to obtain a maximum passive range of motion, to gradually initiate active mobilizations, using active resistance-free, stretching exercises, and to restore the rotator cuff muscle strength.

Phase III of the Neer protocol is aimed at the gradual restoration of the strength and resistance of the shoulder, using stretching and active resistance exercises.

The full achievement of the objectives of a specific phase is mandatory before passing to the next phase, which particularizes the functional rehabilitation treatment.

Although individuals undergoing TSA have less difficulty regaining range of motion initially than those undergoing arthroscopic or open rotator cuff repair, it is still imperative to restore and maintain as much shoulder joint range of motion as possible. It is important to note that although the literature consistently shows significant improvements in strength, range of motion, and functional outcomes after TSA, full range of motion and restoration of full strength are generally not obtained and in some cases not expected (Kasten et al., 2010; Sperling et al., 2008).

Hypothesis

We assessed a young female patient with total shoulder arthroplasty after the excision of a giant-cell tumor of the right proximal humerus as an example for clinical management.

Material and methods

The study was performed in accordance with all current deontological rules. The patient's informed consent was obtained.

Research protocol

Period and place of the research

We report the case of a 24-year-old female patient from a rural area, who presented to the Rehabilitation Hospital in Cluj-Napoca in September 2013, for the restoration of right shoulder mobility after the placement of a total shoulder endoprosthesis.

Subjects and groups

The current disease started insidiously in January 2012, by mixed pain in the right shoulder, of low/moderate intensity, persistent and deep, which was not improved by the administration of analgesics or rest. About 3 months later, in March 2012, the patient observed a progressive limitation of mobility at this level, which is why she presented to the service of Orthopedics of the Baia Mare County Hospital. On radiological examination, a tumor of the right proximal humerus was described. Contrast MRI of the right shoulder was indicated, which evidenced a 4.6/3.6/3.7 cm tumor formation occupying the entire neck of the humerus, with an inhomogeneous structure, with intense contrast uptake, located 7 mm from the scapulohumeral joint in the lower half, which invaded the bone cortex, the subdeltoid bursa, the bicipital sulcus, the insertions of the infraspinatus and supraspinatus muscles, as well as the inner side of the deltoid muscle, with fascial invasion and

mass effect (Fig. 1). The suspicion of a malignant giant-cell tumor was raised. For the confirmation of diagnosis, bone puncture biopsy was indicated, which was performed in June 2012. Anatomic-pathological examination evidenced tissue fragments containing bone tissue, periosteum and reactive bone, as well as images of cell proliferation including a component of mononuclear cells with abundant cytoplasm and a histiocytic appearance with numerous mitoses without atypias, and a component of multinuclear cells, osteogenesis foci apparently located towards the periphery of the tumor proliferations, aneurysmal spaces present inside the proliferation; tumor cells were CD68 positive, CD56 staining was focally positive on cells with osteoblastic appearance, the proliferation index assessed with Ki67 was expressed almost exclusively in the mononuclear component and had a mean value of 30%. The morphological picture, correlated with radiological data, corresponded to a giant-cell tumor of bone.

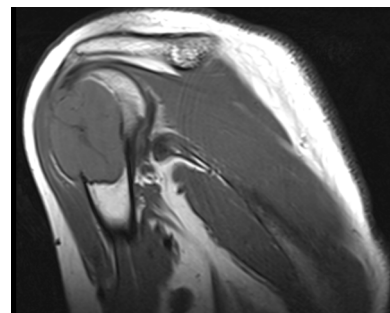


Fig. 1a – T1 sequence

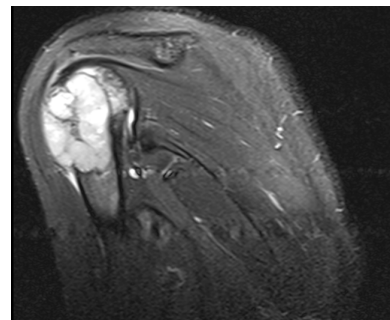


Fig. 1b – T1 sequence, TIRM

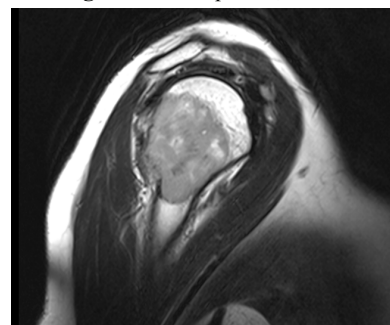


Fig. 1c – T2 sequence

Fig. 1 – Right shoulder MRI, March 2012

In August 2012, surgery was carried out in the Orthopedic Clinic in Munich. The tumor was removed, and metal osteosynthesis with a plate and screws was performed. Postoperative evolution was favorable (Fig. 2). Approximately 6 months after surgery, in January 2013, pain in the right shoulder recurred, and imaging

investigations supported local recurrence. In August 2013, another surgery was performed for the excision of the recurrent tumor, and a titanium total right shoulder endoprosthesis was placed in the same clinic in Munich (Fig. 3).



Fig. 2 – Right shoulder X-Ray (AP), August 2012



Fig. 3 – Right shoulder X-Ray (AP), August 2013

At the first presentation of the patient (September 2013) to the medical rehabilitation service of the Clinical Rehabilitation Hospital Cluj-Napoca, general objective and locomotor system examination was performed, which evidenced: a patient with normal weight (BMI=19 kg/m²), general good status, postoperative scar in the process of healing on the anterior side of the right arm, with minimum painful sensitivity on palpation, hypotonia and hypotrophy

of the right upper limb, and limitation of the mobility of the operated shoulder in all movement planes.

Tests applied

The right shoulder joint was evaluated using goniometry, which showed low range of motion values in all planes, particularly for flexion (5°), abduction (15°) and external rotation (0°). The range of motion values on the first evaluation in September 2013 can be seen in Table I. The patient was also assessed using specific scales: *Constant Shoulder Score*, *Disabilities of the Arm, Shoulder and Hand Score (DASH)*, *Shoulder Pain and Disability Index (SPADI)*. The values obtained on the first evaluation in September 2013 are presented in Table II.

Results and discussion

By correlating clinical and paraclinical data, the objectives of the rehabilitation treatment were formulated: functional rehabilitation of the right shoulder and upper limb, oncologic follow-up, family and socio-professional reintegration, and improvement of the quality of life. Rehabilitation was important because a young adult woman, in full process of developing her personal evolution potential, had lost the optimal function of her dominant upper limb. To the specific rehabilitation objectives, oncologic objectives were added, i.e. monitoring locoregional recurrence and the development of metastases by imaging methods (MRI, CT). Hence, the importance of multidisciplinary collaboration, between the medical rehabilitation specialist, the oncologist, the imaging radiologist, and the family doctor.

The rehabilitation methods used were: kinetotherapy according to phase I of the Neer protocol, associated with water gymnastics in the pool, psychotherapy, occupational therapy, massage and administration of systemic or topical analgesics when needed. It was decided not to use any other form of physiokinetotherapy (e.g. high-frequency short waves), because of the increased recurrence potential of the malignant tumor. The parameters of the rehabilitation program were as follows: 14 days continuous hospitalization, 10 days kinetotherapy (30 minutes twice a day), 10 days water gymnastics in the pool (60 minutes), 10 days sedative massage (20 minutes). At the end of the first rehabilitation program, the range of motion in the right shoulder was significantly improved (Table I), and the

Table I

Range of motion (ROM) of the right shoulder.

Range of motion	September 2013 Admission	September 2013 Discharge	January 2014 Admission	January 2014 Discharge	August 2014 Admission	August 2014 Discharge
Flexion	5°	25°	40°	50°	60°	65°
Extension	30°	30°	30°	35°	40°	40°
Abduction	15°	20°	25°	35°	40°	50°
Internal rotation	10°	15°	20°	20°	20°	20°
External rotation	0°	5°	10°	10°	10°	15°

Table II

Shoulder scores.

Score	September 2013 Admission	September 2013 Discharge	January 2014 Admission	January 2014 Discharge	August 2014 Admission	August 2014 Discharge
Constant Shoulder Score	42	38	35	30	24	22
DASH score	35	33	31.7	30	29.7	29
SPADI score	54%	50%	43%	35%	30%	28%

assessed scores had decreasing values (Table II). The patient was discharged with the following recommendations: continuation of the kinetic program at home, low protein diet, homeopathic treatment, psychotherapy, cognitive-behavioral therapy, and maintenance of contact with the rehabilitation team (consultant doctor, resident doctor, kinesiologist) by phone or Internet.

The patient returned for follow-up after 6 months, in January 2014, with improved range of motion values in the shoulder (the patient reported the daily program of exercises performed at home), and thus, phase II of the Neer protocol could be initiated, following which results were satisfactory (Tables I, II). In August 2014, the patient returned to continue inpatient rehabilitation treatment, and phase III of the Neer protocol was started, with favorable results (Tables I, II). The patient was discharged with the recommendation to continue the kinesiotherapeutic program at home, in order to maintain the obtained values.

Prognosis

Prognosis *ad functionem* is favorable, as there was an increase in the passive and active range of motion, an improvement of muscle strength in the right upper limb, with the amelioration of activities of daily living, as well as family and socio-professional reintegration.

Prognosis *ad vitam* is reserved because of the high risk of local and/or distant recurrence. Also, there are some difficulties in interpreting CT or MRI images due to artefacts generated by the metal endoprosthesis. Long-term monitoring can be done using musculoskeletal ultrasound, which can provide important information on local status.

Particularities of the case

The particularities of the case are the location of the tumor with a starting point in bone that invaded periarticular soft tissues, tumor recurrence requiring surgical reintervention with the placement of a total shoulder endoprosthesis, and the need to adapt kinesiotherapy to the residual anatomic function. The limitations of functional recovery are due to the surgical protocol, which required the resection of the insertions of the rotator cuff muscles in order to remove the head of the humerus, and their subsequent block reinsertion.

Conclusions

1. Physical therapy is an essential determinant of clinical outcome after total shoulder arthroplasty.
2. A graduated rehabilitation program allows patients to obtain a satisfactory overall function of the upper limb.
3. When surgery for the placement of an endoprosthesis is complemented by an early, well-controlled and individualized rehabilitation program, functional prognosis is favorable, and an optimal autonomy of the upper limb, depending on the new anatomical situation, can be obtained.

Conflicts of interests

There are no conflicts of interest.

References

- Becker WT, Dohle J, Bernd L, Braun A, Cserhati M, Enderle A, Hovy L, Matejovsky Z, Szendroi M, Trieb K, Tunn PU. Local recurrence of giant cell tumor of bone after intralesional treatment with and without adjuvant therapy. *The J Bone Joint Surg Am*, 2008;90(5):1060-1067.
- Boudreau S, Boudreau E, Higgins L, Wilcox R. Rehabilitation following reverse total shoulder arthroplasty. *J Orthop and Sports Phys Ther*, 2007;37:734-743.
- Brems JJ. Rehabilitation after total shoulder arthroplasty: current concepts. *Seminars in Arthroplasty*, 2007;18(1):55-65.
- Cahill JB, Cavanaugh JT, Craig EV. Total Shoulder Arthroplasty Rehabilitation. *Techniques in Shoulder & Elbow Surgery*, 2014;15(1):13-17.
- Chakarun CJ, Forrester DM, Gottsegen CJ, Patel DB, White EA, Matcuk GR. Giant cell tumor of bone: review, mimics, and new developments in treatment. *Radiographics*, 2013;33(1):197-211.
- Fornasier VL, Protzner K, Zhang I et al. The prognostic significance of histomorphometry and immunohistochemistry in giant cell tumors of bone. *Hum pathol*, 1996;27(8):754-760.
- Frankle M, Siegal S, Pupello D, Saleem A, Mighell M, Vasey M. The Reverse Shoulder Prosthesis for glenohumeral arthritis associated with severe rotator cuff deficiency. A minimum two-year follow-up study of sixty patients. *J Bone Joint Surg Am*, 2005;87(8):1697-1705.
- Hawkins RJ, Bell RH, Jallay B. Total shoulder arthroplasty. *Clin Orthop Relat Res*, 1989;242:188-194.
- Kasten P, Maier M, Wendy P, Rettig O, Raiss P, Wolf S, Loew M. Can shoulder arthroplasty restore the range of motion in activities of daily living? A prospective 3D video motion analysis study. *J Shoulder Elbow Surg*, 2010;19(2Suppl.):59-65.
- Kay RM, Eckardt JJ, Seeger LL, Mirra JM, Hak DJ. Pulmonary Metastasis of Benign Giant Cell Tumor of Bone: Six Histologically Confirmed Cases, Including One of Spontaneous Regression. *Clin Orthop Relat Res*, 1994;302:219-230.
- Kivioja AH, Blomqvist C, Hietaniemi K, Trovik C, Walloe A, Bauer HC, Jorgensen PH, Bergh P, Follerås G. Cement is recommended in intralesional surgery of giant cell tumors: a Scandinavian Sarcoma Group study of 294 patients followed for a median time of 5 years. *Acta Orthop*, 2008;79(1):86-93.
- Klenke FM, Wenger DE, Inwards CY, Rose PS, Sim FH. Giant cell tumor of bone: risk factors for recurrence. *Clin Orthop Relat Res*, 2011;469(2):591-599.
- Maric M, Bergovec M, Viskovic A, Kolundzic R, Smerdelj M, Orlic D. Treatment and complications in patients with giant cell tumor of bone. *J Bone Joint Surg Br*, 2012;94-B(SUPP XXXVII):257-257.
- Niu X, Zhang Q, Hao L et al. Giant Cell Tumor of the Extremity. *J Bone Joint Surg Am*, 2012;94(5):461-467.
- Rahim A, Tiwari M, Khan G, Gupta G, Sharma S. Total knee arthroplasty with Mesh technique in management of juxta-articular giant cell tumor around the knee. *Int Surg J*, 2015;2(1):47-52.
- Sanchez-Sotelo J. Total shoulder arthroplasty. *Open Orthop J*, 2011;5:106-114. doi: 10.2174/1874325001105010106
- Schajowicz F, Granato DB, McDonald DJ, Sundaram M. Clinical and radiological features of atypical giant cell tumours of bone. *Brit J Radiol*, 1991;64(766):877-889.
- Sperling JW, Kaufman KR, Schleck CD, Cofield RH. A biomechanical analysis of strength and motion following total shoulder arthroplasty. *Int J Shoulder Surg*, 2008;2(1):1-3.
- Szendroi M. Giant-cell tumour of bone. *J Bone Joint Surg Br* 2004;86(1):5-12.