

**ONLINE ARTICLES** 



www.elsevier.com/locate/ymse

CrossMark

# Extra-articular shoulder resections: outcomes of 54 patients

## Andrea Angelini, MD<sup>a</sup>, Andreas F. Mavrogenis, MD<sup>b</sup>, Giulia Trovarelli, MD<sup>a</sup>, Elisa Pala, MD<sup>a</sup>, Pablo Arbelaez, MD<sup>a</sup>, Josè Casanova, MD<sup>c</sup>, Antonio Berizzi, MD<sup>a</sup>, Pietro Ruggieri, MD<sup>a</sup>,\*

<sup>a</sup>Department of Orthopaedics and Orthopaedic Oncology, University of Padova, Padova, Italy <sup>b</sup>First Department of Orthopaedics, National and Kapodistrian University of Athens, School of Medicine, Athens, Greece <sup>c</sup>Department of Orthopaedics, University of Coimbra, Coimbra, Portugal

**Background:** The survival of patients with tumors around the shoulder treated with extra-articular resection, the rates of reconstructions-related complications, and the function of the shoulder cannot be estimated because of limited available data from mainly small published related series and case reports.

**Methods:** We studied 54 patients with tumors around the shoulder treated with extra-articular shoulder resections and proximal humeral megaprosthetic reconstructions from 1985 to 2012. Mean tumor volume was 549 cm<sup>3</sup>, and the mean length of the proximal humeral resection was 110 mm. Mean follow-up was 7.8 years (range, 3-21 years). We evaluated the outcomes (survival, metastases, recurrences, and function) and the survival and complications of the reconstruction.

**Results:** Survival of patients with malignant tumors was 47%, 38%, and 35%, at 5, 10, and 20 years, respectively. Rates for metastasis and local recurrence were 60% and 18.5%, respectively. Survival was significantly higher for patients without metastases at diagnosis, tumor volume <549 cm<sup>3</sup>, and type IV resections. Survival of reconstructions was 56% at 10 years and 48% 20 years. Overall, 19 patients (35.2%) experienced 30 complications (55.5%), the most common being soft tissue failures that required subsequent surgery without, however, implant removal. The mean Musculoskeletal Tumour Society score was 25 points, without any significant difference between the types of extra-articular resections.

**Conclusion:** Tumor stage and volume as well as type of resection are important predictors of survival of patients with malignant tumors around the shoulder. Survival of the reconstructions is satisfactory; nevertheless, the complication rate is high. The Musculoskeletal Tumour Society score is similar with respect to the type of resection.

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

 $\ensuremath{\mathbb{C}}$  2017 Journal of Shoulder and Elbow Surgery Board of Trustees. All rights reserved.

Keywords: Sarcoma; shoulder; resection; Tikhoff-Linberg; extra-articular; outcome

\*Reprint requests: Pietro Ruggieri, MD, University of Padova, Via Giustiniani 3, 35128, Padova, Italy.

E-mail address: pietro.ruggieri@unipd.it (P. Ruggieri).

Tumors around the shoulder account for approximately onethird of all tumors<sup>10,28,39</sup>; the proximal humerus, scapula, and clavicle are the most common sites.<sup>47</sup> Until the 1970s, forequarter amputations and shoulder disarticulations were the treatment of choice for most patients with malignant tumors of the shoulder.<sup>47</sup> Current advances in imaging, surgery, and

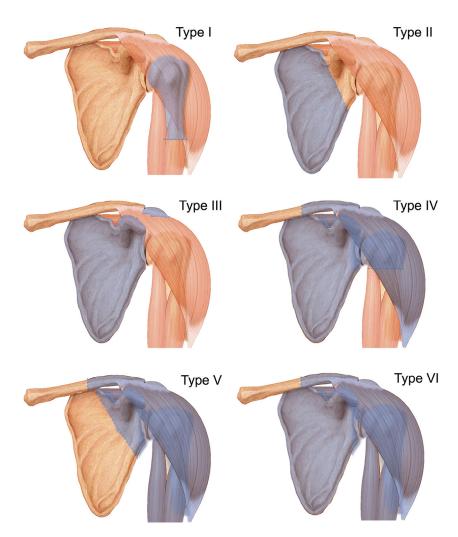
1058-2746/\$ - see front matter © 2017 Journal of Shoulder and Elbow Surgery Board of Trustees. All rights reserved. https://doi.org/10.1016/j.jse.2017.04.019

This study was approved by the Institutional Review Board/Ethics Committee of the authors' institutions.

adjuvant treatments have enabled limb salvage surgery for 80% to 95% of these patients.<sup>2,47</sup> A forequarter amputation is indicated for tumors involving the neurovascular bundle, for recurrent tumors, when bypass surgery cannot be performed or wide repeat resection is not feasible, and for failed limb-salvage resections and reconstructions.<sup>8,23-25,28,30,31,33</sup>

Partial scapulectomy was first reported by Liston in 1819 for an ossified aneurysmal tumor. Most shoulder girdle resections since then were done for low-grade tumors of the scapula and periscapular soft-tissue sarcomas.<sup>27-29</sup> Wide (microscopically negative) resection of a sarcoma that extends to the shoulder joint requires an extra-articular glenohumeral joint resection.<sup>8,23-25,28,30,31,33</sup> Total scapulectomy and extraarticular resection of the glenohumeral joint by an osteotomy inferior to the glenohumeral capsule indicates the Tikhoff-Linberg resection. After the initial description of the Tikhoff-Linberg resection for osteosarcoma and Ewing sarcoma of the proximal humerus and scapula in the 1980s, modifications of the typical procedure have been developed.<sup>1,3,9,11,14,15,17,21,27,29,32,34-36,38,42-46</sup> The typical Tikhoff-Linberg resection does not preserve the deltoid or trapezius muscles; however, to provide adequate soft tissue coverage for the scapular prosthesis, these muscles must be retained (modified Tikhoff-Linberg resection).<sup>24,27,29,32</sup> Another important modification has been the excision of only the lateral part of the scapula, whenever possible.<sup>8,28</sup>

Malawer et al<sup>28</sup> described 3 techniques for intra-articular shoulder resections (types I, II, and III) and 3 for extraarticular (Tikhoff-Linberg) shoulder resections (Fig. 1). After extra-articular resection, reconstruction options include megaprosthetic or osteoarticular allografts, or both.<sup>6-8,19,40</sup> The extent of tumor resection and remaining muscles for



**Figure 1** An illustration shows the types of intra-articular (types I, II, and III) and extra-articular (Tikhoff-Linberg) shoulder resections (types IV, V, and VI) according to Malawer et al.<sup>28</sup> Type I includes intra-articular proximal humeral resection, type II includes partial scapular resection, type III includes intra-articular total scapulectomy, type IV includes extra-articular total scapulectomy and humeral head resection, type V includes extra-articular total scapulectomy and proximal humeral resection. Each type is divided into a subtype A or B depending on whether the abductor muscles are retained (subtype A) or resected (subtype B) with the tumor specimen.

reconstruction indicate the degree of shoulder motion and function.  $^{\mbox{\tiny 28}}$ 

The survival of patients with tumors around the shoulder girdle treated with extra-articular resection, the rates of reconstruction-related complications, and the function of the shoulder cannot be estimated because of limited available data from mainly small published related series and case reports.<sup>8,15,16,22,26,28,30-32,42</sup> Therefore, to enhance the literature, we performed this study of patients with tumors around the shoulder treated with extra-articular shoulder resection and megaprosthetic reconstructions to evaluate the outcome of the patients (survival, metastases, recurrences, and function) and the survival and complications of the reconstructions.

#### Materials and methods

We retrospectively studied the files of 54 patients with aggressive benign and malignant bone (50 patients) and soft-tissue (4 patients) tumors around the shoulder girdle treated with extra-articular shoulder resection and megaprosthetic reconstruction from January 1985 to August 2012. There were 34 male and 20 female patients with a mean age of 46 years (range, 15-84 years). Tumor location was the proximal humerus in 33 patients, the scapula in 17, and the soft tissue around the shoulder joint in 4 (Table I). The most common present-

Variable	Patients
	No. (%)
Gender	
Male	34 (63)
Female	20 (37)
Tumor location	
Proximal humerus	33 (61)
Scapula	17 (31)
Soft tissue around the shoulder	4 (7)
Diagnosis	
Chondrosarcoma	22 (41)
Osteosarcoma	15 (28)
Metastasis	6 (11)
Synovial sarcoma	2 (4)
Angiosarcoma	1 (1.8)
Fibrosarcoma	1 (1.8)
Pleomorphic sarcoma	1 (1.8)
Lymphoma	1 (1.8)
Liposarcoma	1 (1.8)
Ewing sarcoma	1 (1.8)
Giant cell tumor	1 (1.8)
Aggressive fibromatosis	1 (1.8)
Chondroblastoma	1 (1.8)
Stage <sup>*</sup>	
Stage 3	3 (5.5)
Stage IIB	40 (74)
Stage III	11 (20)

\* Enneking staging system for musculoskeletal tumors.<sup>13</sup> (Stage 3 refers to the three patients with benign bone tumors. Stage IIB and III refer to the patients with malignant tumors.) ing symptom was pain with swelling and functional limitation of the shoulder; 3 patients presented with a pathologic fracture.

Indications for extra-articular resection were (1) tumors of the scapula, proximal humerus, lateral clavicle, or periscapular soft tissue with invasion of the subchondral bone, joint capsule, synovial membrane, or the entire joint as evident on staging imaging (43 patients), and (2) patients with malignant tumors and lung metastases who refused a forequarter amputation (11 patients). Contraindication for extra-articular resection was tumor involvement of the axillary neurovascular bundle. The mean follow-up was 7.8 years (range, 3-21 years). Follow-up was 3 to 5 years in 35 patients, 5 to 10 years in 7, 10 to 20 years in 9, and more than 20 years in 3. No patient was lost to follow-up. No patient was specifically recalled for the purpose of this study. All data were retrieved from patient files and surgical reports. At admission, all patients gave written informed consent for their data to be included in this study.

All patients had preoperative staging<sup>13</sup> with radiographs and magnetic resonance imaging (MRI) of the shoulder, computed tomography (CT) of the chest, and bone scan, followed by CT-guided needle biopsy for histologic diagnosis of their tumors. Adjuvant treatments, including chemotherapy or radiotherapy, or both, were administered to patients with malignant tumors according to the histology and grade of the tumor.

Tumor volume was measured on coronal, transverse, and sagittal MRIs. The maximum height, width, and depth were recorded, and the volume was calculated using the formula of an ellipsoid mass volume (tumor volume =  $[\pi/6] \times$  height × width × depth).<sup>41</sup> The mean tumor volume of the patients in this series was 549 cm<sup>3</sup> (range, 9-2571 cm<sup>3</sup>).

Extra-articular shoulder resection consisted of a type IV resection in 14 patients, a type V resection in 21 (including the 3 patients with aggressive benign tumors), and a type VI resection in 19.<sup>28</sup> Resection types were further classified as typical Tikhoff-Linberg resections (Malawer types IV and VI) and modified Tikhoff-Linberg resections (Malawer type V). A modified Tikhoff-Linberg resection was indicated when the tumor abutted or involved the shoulder joint, without or with minimal invasion of the glenoid. The mean length of proximal humeral resection was 110 mm (range, 50-230 mm).

In all patients, the deltoid muscle and rotator cuff was resected en bloc with the tumor specimen, aiming for wide (microscopically negative) resection margins; if a small cuff of deltoid muscle was spared, it was used as a soft tissue envelope and was not reattached. The radial nerve was sacrificed in 6 patients, and the musculocutaneous and median nerves were sacrificed in 1 patient each. Reconstruction after extra-articular resection was done in all patients with a modular proximal humerus megaprosthesis (cemented in 33 patients and cementless in 21) that was sutured and suspended from the remaining clavicle in 33 patients or the scapula in 21 patients with nonabsorbable No. 5 Ethibond (Ethicon, Somerville, NJ, USA) transosseous sutures (47 patients) and an artificial mesh (7 patients). The scapula was not replaced in any of the patients. After megaprosthetic reconstruction, the remaining muscles, including the long head of the biceps muscle, pectoralis major and minor muscles, and triceps muscle were sutured and approximated to cover the implants and restore stability.

Histologic examination of the excised tumor specimens showed wide or marginal resection margins (microscopically negative) in 47 patients, and marginal (3 patients) or intralesional (1 patient) resection margins (microscopically positive) in 4 patients.

Postoperative follow-up evaluation was done every 3 months for the first 2 years, every 6 months for the next 3 years and annually thereafter for the patients with malignant tumors. The follow-up evaluation was done every 6 months for the first 3 years and annually thereafter for the patients with aggressive benign tumors. The followup evaluation included clinical examination, radiographs, MRI or CT scan, or both, of the shoulder. A chest CT was done every 6 months for the first 3 years and then annually for the patients with malignant tumors.

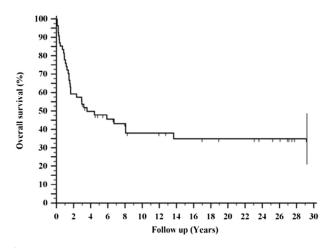
The outcome of the patients (survival, local recurrences, metastases, and function) and the survival and complications of the reconstructions were evaluated at each follow-up examination and at the last follow-up for the purpose of this study. The patients were classified as no evidence of disease (NED), alive with disease (AWD), or dead of the disease (DWD).

Categoric variables are expressed as the number of occurrences and percentage of the total patients in a category. Survival was analyzed using the Kaplan-Meier analysis.<sup>20</sup> The curves were compared with a log-rank test,<sup>37</sup> where the starting point was surgery and the end point was the occurrence of death for the patients or revision and forequarter amputation for the megaprostheses. Patients who died with their implants in place were censored.

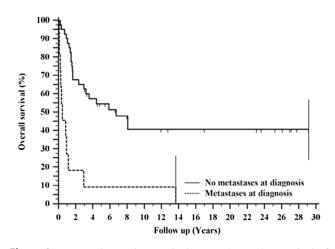
Multivariate analysis was performed to examine independent risk factors for survival using the Cox regression model with a stepwise forward procedure,<sup>20</sup> and tumor stage for malignant tumors (Enneking stage<sup>13</sup> IIB vs. stage III), tumor volume (<549 cm<sup>3</sup> vs. >549 cm<sup>3</sup>, which was the mean tumor volume of the patients in this series), and resection type (typical vs. modified Tikhoff-Linberg resection). Reconstruction-related complications were classified according to Henderson et al18 into failures of the soft tissues around the implant (type 1), aseptic loosening of the implant (type 2), structural fracture (type 3), infection (type 4), and tumor recurrences (type 5). Function was compared to the contralateral upper extremity and evaluated with the Musculoskeletal Tumor Society (MSTS) functional rating system,<sup>12</sup> which is the most commonly used system for evaluation of function in tumor patients that enables results between studies to be compared. Data were recorded in an Excel 2010 spreadsheet (Microsoft Corporation, Redmond, WA, USA) and analyzed using MedCalc Software 11.1 (MedCalc Software, Mariakerke, Belgium).

#### Results

Survival of the patients with malignant tumors was 47%, 38%, and 35%, at 5, 10, and 20 years, respectively (Fig. 2). At the last follow-up, 16 patients had NED, 2 patients were AWD, and 33 patients were DWD. The 3 patients with benign tumors had NED. Survival of the patients with malignant tumors was statistically significantly higher for the patients without metastases at diagnosis than for the patients with metastases at diagnosis (P < .0001, Fig. 3) and for the patients with tumor volume <549 cm<sup>3</sup> compared with the patients with tumor volume >549 cm<sup>3</sup> (P = .0011, Fig. 4). Survival of the patients with malignant tumors was not statistically significantly different with respect to the histology of the primary tumors (P = .3289) and the type of extra-articular resection (typical or modified Tikhoff-Linberg resection, P = .4723). By direct comparison of the survival with respect to the types of extraarticular resection, survival was statistically significantly higher for the patients who had a type IV compared to a type V



**Figure 2** A Kaplan-Meier survival curve shows the survival of patients with malignant tumors was 47%, 38%, and 35% at 5, 10, and 20 years, respectively.

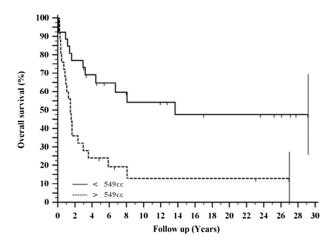


**Figure 3** A Kaplan-Meier survival curve shows the survival of patients with malignant tumors was statistically significantly higher for patients without metastases at diagnosis compared with the survival of patients with metastases at diagnosis (P < .0001).

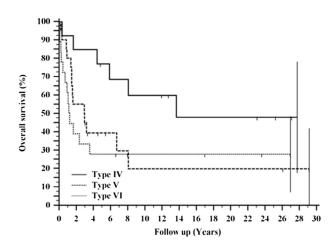
(P = .0334) or a type VI (P = .0359) resection (Fig. 5). Multivariate Cox regression analysis showed that tumor stage and volume as well as type of resection were important predictors of survival of the patients with malignant tumors (Table II).

The metastasis rate of the 40 patients with malignant tumors without a metastasis at diagnosis (stage IIB) was 60%, and 24 of these patients experienced a lung metastasis at a mean of 1.5 years (range, 2.5 months-3.5 years) after diagnosis and treatment. These patients were treated with metastasectomy, radiotherapy, chemotherapy, or a combination. At the last follow-up, 3 of the 24 patients with malignant tumors without a metastasis at diagnosis (stage IIB) were NED and 21 were DWD. All patients with malignant tumors with a metastasis at diagnosis (stage III) were DWD.

Survival of the reconstructions was 56% at 10 years and 48% at 20 years (Fig. 6). Overall, 19 patients (35.2%) experienced 30 complications (55.5%) at a mean of 1.4 years (range, 1 month-5 years) after treatment (Table III). Soft tissue



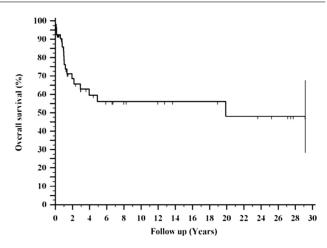
**Figure 4** A Kaplan-Meier survival curve shows the survival of patients with tumor volume  $<549 \text{ cm}^3$  was statistically significantly higher compared with the survival of patients with a tumor volume  $>549 \text{ cm}^3$  (P = .0011).



**Figure 5** A Kaplan-Meier survival curve shows the survival of patients who had a type IV resection was statistically significantly higher compared with patients who had a type V (P = .0334) or a type IV (P = .0359) resection.

<b>Table II</b> Multivariate Cox regression analysis: tumor stage and volume and type of resection were important predictors of survival of the patients with malignant tumors included in this series					
Covariate	OR	95% CI	P value		
Stage IIB vs. stage III Typical vs. modified Tikhoff-Linberg resection	5.99 3.16	2.1750-16.5058 1.3749-7.2688	.0005 .0070		
Tumor volume (<549 cm <sup>3</sup> vs. >549 cm <sup>3</sup> )	6.92	2.8621-16.7619	<.0001		
<i>CL</i> confidence interval: <i>OR</i> : 0	dds ratio				

complications occurred in 14 patients, including failure of suture suspension and dislocation of the megaprosthesis in 8 patients and wound dehiscence in 6. Failure of suture suspension and dislocation of the megaprosthesis were treated



**Figure 6** A Kaplan-Meier survival curve shows the survival of the reconstructions was 56% at 10 years and 48% at 20 years.

**Table III**Overall complications after extra-articular resections in this series

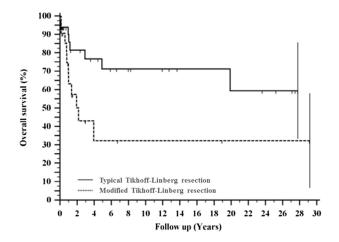
Complications	Type of resection, No. (%)		
	Type IV	Type V	Type VI
Type 1 (soft tissue failure)	2 (7.1)	7 (23.8)	5 (21)
Type 2 (aseptic loosening)	_	_	1 (5.3)
Type 3 (structural failure)	-	2 (9.5)	1 (5.3)
Type 4 (infection)	-	2 (9.5)	-
Type 5 (local recurrence)	3 (21.4)	6 (28.6)	1 (5.3)

with reattachment of the megaprosthesis with sutures (7 patients) and a polyethylene terephthalate synthetic tube (1 patient).

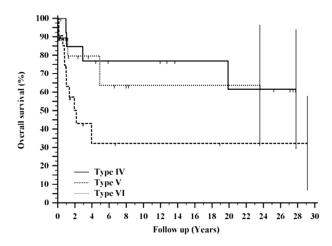
Wound dehiscence was treated successfully with débridement only in 4 patients and with débridement and muscle flap coverage in 2 patients. Two patients experienced a deep infection and were treated with a 2-stage revision surgery (1 patient) and a forequarter amputation (1 patient). Aseptic loosening occurred in 1 patient and was treated with revision of the megaprosthesis. Periprosthetic fractures in 2 patients were treated with open reduction and plate and screw fixation, and another patient experienced breakage of the stem of the megaprosthesis and was treated with revision of the megaprosthesis.

The rate of local recurrence of the patients in this series was 18.5%: 3 of 14 patients (21.4%) with a type IV resection, 6 of 21 patients (28.5%) with a type V resection, and 1 of 19 patients (5.3%) with a type VI resection experienced a local recurrence at a mean time of 3.4 years (range, 7 months-19 years) after diagnosis and treatment. These patients were treated with a forequarter amputation (6 patients), repeat resection (3 patients), and palliative radiotherapy (1 patient). At the last follow-up, 7 patients were NED after treatment of their recurrence, and a forequarter amputation was performed in 2 patients because of repeat recurrence after treatment.

Survival to failure of the reconstructions was statistically significantly higher for the patients treated with the typical



**Figure 7** A Kaplan-Meier survival curve shows the survival to failure of the reconstructions was statistically significantly higher for patients treated with the typical Tikhoff-Linberg resection compared with those treated with the modified Tikhoff-Linberg resection (P = .0212).



**Figure 8** A Kaplan-Meier survival curve shows the survival to failure of the reconstructions was statistically significantly higher for patients who had a type IV resection compared with those who had a type V (P = .0313) or type VI resection (P = .6330), without any difference between the type V and type VI resections (P = .1300).

Tikhoff-Linberg resection compared with those treated with the modified Tikhoff-Linberg resection (P = .0212; Fig. 7), and for the patients treated with the type IV compared with the type V (P = .0313) and type VI resection (P = .6330), without any difference between the type V and type VI resections (P = .1300; Fig. 8). Survival to local recurrence of the resections and megaprosthetic reconstructions was not different with respect to the types of extra-articular resection (P = .1220).

Shoulder motion was restricted compared with the contralateral shoulder in all patients. Function of the elbow, forearm, and hand was similar to the contralateral upper extremity in all except the 8 patients whose ipsilateral radial, musculocutaneous, and median nerves were resected with the tumor specimen. The overall mean MSTS score was 25 points (range, 17-30 points) and was 25 points in patients who had a type IV resection, 26 points in patients who had a type V resection, and 24 points in patients who had a type VI resection, without any statistically significant difference with respect to the type of the resection (P = .736).

#### Discussion

The Tikhoff-Linberg extra-articular shoulder resection is a feasible surgical treatment to forequarter amputation and shoulder disarticulation for malignant and aggressive benign tumors of the shoulder.<sup>7,8,10,15,16,19,22-25,28,30,31,33,47</sup> Baumann<sup>4</sup> in 1914 reported excision of the scapula, the head of the humerus, the lateral one-third of the clavicle, and the surrounding soft tissue for a sarcoma of the scapula. Tikhoff and Baumann performed 3 such operations between 1908 and 1913; Tikhoff was named as the originator of the "triple-bone resection technique" that became known as the Tikhoff-Linberg resection after the English publication in 1926.<sup>24</sup> In 1991, Malawer et al<sup>28</sup> proposed a 6-type classification system of shoulder resections based on the bony segments involved and their relationship to the glenohumeral joint, and 2 subtypes (A and B) according to whether the shoulder abductor mechanism was preserved (subtype A) or resected (subtype B).<sup>27,28</sup> Since then, a variety of techniques and modifications of shoulder girdle resections have been described. <sup>1,3,5,8,9,11,14,15,17,21,23-25,27-36,38,42-47</sup> However, the related literature is limited by small series and case reports of variable techniques<sup>8,15,16,22,26,28,30-32,42</sup>; their results cannot be summarized to draw important conclusions regarding the treatment approach and the outcome of the patients and reconstructions after shoulder resections.

Therefore, to enhance the literature, we performed this study of a relatively large number of patients with tumors around the shoulder girdle treated with extra-articular resection aiming to evaluate the outcome of the patients and the reconstructions. Our results showed (1) a survival rate of 47% to 35% at 5 to 20 years of the patients with malignant tumors, which was higher for the patients without metastases at diagnosis, tumor volume  $<549 \text{ cm}^3$ , and type IV resections, (2) a metastasis rate of 60% for the patients with malignant tumors around the shoulder, (3) a local recurrence of 18.5%, which was not different with respect to the types of extra-articular resection, (4) a complications rate of 55.5%, and (5) a mean MSTS score of 25 points, which was also not different with respect to the types of extra-articular resection. Type IV resections were associated with a significantly higher survival of the patients and the reconstructions. The former should probably be attributed to the lower tumor burden for these patients, and the latter should probably be attributed to retention of a large part of the humerus for a proximal humeral megaprosthesis, with or without reconstruction of the scapula to be performed.

Marcove et al<sup>31</sup> reported 17 patients treated with the typical Tikhoff-Linberg resection. At a follow-up ranging from 1 to 33 months, 9 patients were NED and 1 patient

was AWD. Local recurrence occurred only in 1 patient at the cervical spine, and the patient eventually died of his disease. Guerra et al<sup>15</sup> reported 18 patients treated with the typical Tikhoff-Linberg resection and 3 patients treated with the modified Tikhoff-Linberg resection. At a mean followup of 12 months (range, 3-36 months), 16 patients were NED, 3 patients were AWD, and 2 patients were DWD. Local recurrence occurred only in 1 patient after a marginal margins resection.

Capanna et al<sup>8</sup> compared 12 patients treated with the typical Tikhoff-Linberg resection and 12 patients treated with the modified Tikhoff-Linberg resection. At a mean follow-up of 22.5 months, 14 patients were NED, 1 was NED after treatment for a metastasis, 2 were AWD, and 7 were DWD, without any difference between the 2 types of resection. These authors concluded that for tumors of the proximal humerus, the typical Tikhoff-Linberg resection is indicated when preoperative imaging studies show macroscopic involvement of the shoulder joint with invasion of the scapula, whereas the modified resection is indicated in cases of invasion of the joint capsule without involvement of the glenoid.<sup>8</sup>

Voggenreiter et al<sup>42</sup> reported 19 patients treated with a Tikhoff-Linberg resection. At a mean follow-up of 6.3 years, 12 patients were NED, 1 was AWD (lung metastasis and local recurrence), and 7 were DWD.

In the present study at the last follow-up, 16 patients were NED, 2 patients were AWD, and 33 patients were DWD; 24 patients (60%) experienced a metastasis, 10 patients (18.5%) experienced a local recurrence, and 19 patients (35.2%) experienced complications. We concur with previous reports<sup>8,15</sup> that the modified resection is indicated when the tumor abuts or involves the shoulder joint without or with minimal invasion of the glenoid. However, our results showed that survival of the patients, risk for metastasis and local recurrence, and function were not statistically significantly different with respect to the type of resections. Only the survival of the reconstructions was statistically significantly higher after the typical Tikhoff-Linberg resection compared with the modified technique; probably, trying to retain muscles for soft tissue coverage of a proximal humerus reconstruction and to excise only the lateral part of the scapula with subsequent partial scapula or glenoid reconstruction may increase the risk of related complications.

The most commonly reported complications after extraarticular shoulder resections are soft tissue compromises,<sup>31,42</sup> which do not usually require revision surgery with implant removal. Marcove et al<sup>31</sup> reported 4 patients with wound dehiscence, and only 1 required surgical revision with a splitthickness skin graft. Voggenreiter et al<sup>42</sup> reported 3 patients with wound dehiscence that required surgical revision and 2 patients with dislocation of the megaprosthesis caused by a polyethylene terephthalate synthetic tube rupture. In the present series, soft tissue compromises, including wound dehiscence and dislocations, were also the most common complications, which required subsequent surgery, without however revision of the implant. Complications that require removal of the megaprosthesis are less common.<sup>8,16,22,42</sup> Capanna et al<sup>8</sup> reported 4 patients who experienced a deep infection, and Voggenreiter et al<sup>42</sup> reported 1 patient who experienced a deep infection and 2 patients who experienced implant breakage. In the present series, we observed a similar rate of complications as the previous reports. Most complications can be treated successfully without implants removal. Revision of the megaprosthesis is usually required for patients who experience deep infection, breakage, or aseptic loosening of their implants.

Extra-articular shoulder resections allow for almost normal function of the hand and forearm, with reasonable function of the elbow.<sup>8,10,15,16,19,22-25,28,30,31,33,37</sup> Voggenreiter et al<sup>42</sup> reported a mean MSTS score of 72%, and Yang et al<sup>47</sup> reported a mean MSTS score of 67% after type IV resections, 73% after type V resections, and 40% after type VI resections. In a case report, Mackinnon et al<sup>26</sup> reported a patient with a flail shoulder who was able to lift a 25 kg weight by flexing his elbow. Hahn et al<sup>16</sup> reported that function of the shoulder is poor after extra-articular shoulder resections when the humerus is resected at about more than half of its length; in contrast, function of the shoulder is good when the humerus is resected at approximately its anatomic neck. In the present series, we observed a mean MSTS score of 25 points (range, 17-30 points), which is considered good, without a statistically significant difference with respect to the type of the resection (typical or modified).

This study has 3 limitations. First, it is retrospective and nonrandomized, subjecting it to potential recall and selection biases; however, the rarity of the disease would make a prospective study practically unfeasible.

Second, the study period spanned more than 3 decades and included patients treated for variable musculoskeletal tumors with a variety of adjuvant treatments by different surgeons and medical oncologists. During the study period, obviously, diagnostic and treatment approaches have changed; therefore, the possibility of confounding variables is important. Again, considering the rarity of the type of treatment, concentrating a study on patients treated over a short period of time would not be possible. We acknowledge that this adds to the heterogeneity of our sample size and concur that the patients with malignant tumors are at risk for distant metastases, higher risk of cancer, and implants-related complications resulting from by adjuvant treatments. However, we aimed to include all of our patients with tumors around the shoulder girdle, including aggressive benign and malignancies treated with extra-articular shoulder resections, aiming to increase the sample size and draw useful conclusions for this treatment approach. In addition, we studied only patients treated after 1985 to have a more homogeneous series after introduction of chemotherapy and with a minimum follow-up of 3 years. We believe that in this setting, our results are valid.

Third, one may consider a survivorship bias with respect to the survival of the patients and the survival of the reconstructions. This is a logical mistake when concentrating on survivors of a process and omitting those who did not survive because of their lack of visibility. We also acknowledge this limitation. To avoid this bias and statistical error in this study, survivorship was adjusted according to the follow-up time for each patient, follow-up time (mean, range, and minimum) was clarified, and data from all patients were analyzed from the patients' files and reports. In this setting, we believe that our results are useful and valid.

### Conclusions

The survival of patients with malignant tumors around the shoulder girdle is higher for patients without metastases at diagnosis, tumor volume <549 cm<sup>3</sup>, and type IV resections. Local recurrence is not different between the typical and the modified resections. Survival of the reconstructions is satisfactory; nevertheless, the complication rate is high. Soft tissue complications are the most common, without, however, requiring implants removal. Shoulder motion is restricted, but the MSTS score is not different with respect to the type of resection.

#### 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

- Abouljoud MS, Nathanson SD. Use of polypropylene mesh to stabilize the "floating humerus" after total scapulectomy. Surgery 1991;110:905-8.
- Bacci G, Ferrari S, Comandone A, Zanone A, Ruggieri P, Longhi A, et al. Neoadjuvant chemotherapy for Ewing's sarcoma of bone in patients older than thirty-nine years. Acta Oncol 2000;39:111-6.
- Baran O, Havitçioğlu H. Extremity saving surgery and reconstruction for tumors of the scapula. Acta Orthop Traumatol Turc 2007;41:195-201.
- Baumann PK. Resection of the upper extremity in the region of the shoulder joint. Khirurgh Arkh Velyaminova 1914;30:145.
- Bickels J, Wittig JC, Kollender Y, Kellar-Graney K, Meller I, Malawer MM. Limb-sparing resections of the shoulder girdle. J Am Coll Surg 2002;194:422-35. http://dx.doi.org/10.1016/S1072-7515(02)01124-9
- 6. Capanna R, Mapelli S, Ruggieri P, Biagini R, Ferruzzi A, Zucchi V, et al. Resection of the proximal humerus and I.O.R. modular prosthesis in the treatment of metastatic lesions. Ital J Orthop Traumatol 1988;14:143-8.
- Capanna R, van Horn JR, Biagini R, Ruggieri P, Bettelli G, Sola G, et al. A humeral modular prostheses for bone tumour surgery: a study of 56 cases. Int Orthop 1986;10:231-8.
- Capanna R, van Horn JR, Biagini R, Ruggieri P, Ferruzzi A, Campanacci M. The Tikhoff-Linberg procedure for bone tumors of the proximal humerus: the classical "extensive" technique versus a modified "transglenoid" resection. Arch Orthop Trauma Surg 1990;109:63-7.
- Clarke A, Dewnany G, Neumann L, Wallace WA. Glenothoracic fusion. An adjunct to radical scapulectomy. J Bone Joint Surg Br 2004;86:531-5. http://dx.doi.org/10.1302/0301-620X.86B4.14367

- Creighton JJ, Peimer CA, Mindell ER, Boone DC, Karakousis CP, Douglass HO. Primary malignant tumors of the upper extremity: retrospective analysis of one hundred twenty-six cases. J Hand Surg Am 1985;10:805-14.
- Damron TA, Rock MG, O'Connor MI, Johnson M, An KN, Pritchard DJ, et al. Functional laboratory assessment after oncologic shoulder joint resections. Clin Orthop Relat Res 1998;(348):124-34.
- Enneking WF, Dunham W, Gebhardt MC, Malawar M, Pritchard DJ. A system for the functional evaluation of reconstructive procedures after surgical treatment of tumors of the musculoskeletal system. Clin Orthop Relat Res 1993;(286):241-6.
- Enneking WF, Spanier SS, Goodman MA. A system for the surgical staging of musculoskeletal sarcoma. Clin Orthop Relat Res 1980;(153):106-20.
- Francis KC, Worcester JN. Radical resection for tumors of the shoulder with preservation of a functional extremity. J Bone Joint Surg Am 1962;44:1423-30.
- Guerra A, Capanna R, Biagini R, Ruggieri P, Campanacci M. Extraarticular resection of the shoulder (Tikhoff-Linberg). Ital J Orthop Traumatol 1985;11:151-7.
- Hahn SB, Kim NH, Choi NH. Treatment of bone tumors around the shoulder joint by the Tikhoff-Linberg procedure. Yonsei Med J 1990;31:110-22.
- Ham SJ, Hoekstra HJ, Eisma WH, Schraffordt Koops H, Oldhoff J. The Tikhoff-Linberg procedure in the treatment of sarcomas of the shoulder girdle. J Surg Oncol 1993;53:71-7.
- Henderson ER, Groundland JS, Pala E, Dennis JA, Wooten R, Cheong D, et al. Failure mode classification for tumor endoprostheses: retrospective review of five institutions and a literature review. J Bone Joint Surg Am 2011;93:418-29. http://dx.doi.org/10.2106/JBJS.J.00834
- Jensen KL, Johnston JO. Proximal humeral reconstruction after excision of a primary sarcoma. Clin Orthop Relat Res 1995;(311):164-75.
- Kaplan EL, Meier P. Nonparametric estimation from incomplete observations. J Am Stat Assoc 1958;53:457-81.
- Kiss J, Sztrinkai G, Antal I, Kiss J, Szendroi M. Functional results and quality of life after shoulder girdle resections in musculoskeletal tumors. J Shoulder Elbow Surg 2007;16:273-9. http://dx.doi.org/10.1016/ j.jse.2006.08.011
- Kofránek I, Matejovský Z. The Tikhoff-Linberg procedure. Acta Chir Orthop Traumatol Cech 1999;66:76-81.
- Kumar VP, Satku SK, Mitra AK, Pho RW. Function following limb salvage for primary tumors of the shoulder girdle. 10 patients followed 4 (1-11) years. Acta Orthop Scand 1994;65:55-61.
- Linberg BE. Interscapulo-thoracic resection for malignant tumors of the shoulder girdle region. J Bone Joint Surg Am 1928;10:344.
- Machson NJ. Modification of the interscapulo-thoracic resection (Tichow-Linberg's operation). Beitr Orthop Traumatol 1968;15:212-5.
- Mackinnon J, Aziz T, Dixon JH. Interscapulothoracic resection. A case report and review of the literature. J Bone Joint Surg Br 1988;70:791-4.
- Malawer MM. Tumors of the shoulder girdle. Technique of resection and description of a surgical classification. Orthop Clin North Am 1991;22:7-35.
- Malawer MM, Meller I, Dunham WK. A new surgical classification system for shoulder-girdle resections. Analysis of 38 patients. Clin Orthop Relat Res 1991;(267):33-44.
- Malawer MM, Sugarbaker PH. Musculoskeletal cancer surgery. Norwell, MA: Kluwer Academic Publishers; 2001 ISBN: 0792363949, 9780792363941.
- Malawer MM, Sugarbaker PH, Lampert M, Baker AR, Gerber NL. The Tikhoff-Linberg procedure: report of ten patients and presentation of a modified technique for tumors of the proximal humerus. Surgery 1985;97:518-28.
- Marcove RC, Lewis MM, Huvos AG. En bloc upper humeral interscapulo-thoracic resection. The Tikhoff-Linberg procedure. Clin Orthop Relat Res 1977;(124):219-28.
- Mavrogenis AF, Mastorakos DP, Triantafyllopoulos G, Sakellariou VI, Galanis EC, Papagelopoulos PJ. Total scapulectomy and constrained

reverse total shoulder reconstruction for a Ewing's sarcoma. J Surg Oncol 2009;100:611-5. http://dx.doi.org/10.1002/jso.21340

- Meller I, Bickels J, Kollender Y, Ovadia D, Oren R, Mozes M. Malignant bone and soft tissue tumors of the shoulder girdle. A retrospective analysis of 30 operated cases. Acta Orthop Scand 1997;68:374-80.
- Mnaymneh WA, Temple HT, Malinin TI. Allograft reconstruction after resection of malignant tumors of the scapula. Clin Orthop Relat Res 2002;(405):223-9.
- Nakamura S, Kusuzaki K, Murata H, Takeshita H, Hirata M, Hashiguchi S, et al. Clinical outcome of total scapulectomy in 10 patients with primary malignant bone and soft-tissue tumors. J Surg Oncol 1999;72:130-5.
- O'Connor MI, Sim FH, Chao EY. Limb salvage for neoplasms of the shoulder girdle. Intermediate reconstructive and functional results. J Bone Joint Surg Am 1996;78:1872-88.
- Petrie A. Statistics in orthopaedic papers. J Bone Joint Surg Br 2006;88:1121-36. http://dx.doi.org/10.1302/0301-620X.88B9.17896
- Pritsch T, Bickels J, Wu CC, Squires MH, Malawer MM. Is scapular endoprosthesis functionally superior to humeral suspension? Clin Orthop Relat Res 2007;456:188-95.
- Rosenberg SA, Suit FD, Baker LH. Sarcomas of soft tissue. In: Devita VT, Hellman S, Rosenberg SA, editors. Cancer: principles and practice of oncology. 2nd ed. Philadelphia: JB Lippincott; 1985. p. 1243-93.
- Ruggieri P, Mavrogenis AF, Guerra G, Mercuri M. Preliminary results after reconstruction of bony defects of the proximal humerus with an

allograft-resurfacing composite. J Bone Joint Surg Br 2011;93:1098-103. http://dx.doi.org/10.1302/0301-620X.93B8.26011

- Shin KH, Moon SH, Suh JS, Yang WI. Tumor volume change as predictor of chemotherapeutic response in osteosarcoma. Clin Orthop Relat Res 2000;(376):200-8.
- Voggenreiter G, Assenmacher S, Schmit-Neuerburg KP. Tikhoff-Linberg procedure for bone and soft tissue tumors of the shoulder girdle. Arch Surg 1999;134:252-7.
- Volpe CM, Pell M, Doerr RJ, Karakousis CP. Radical scapulectomy with limb salvage for shoulder girdle soft tissue sarcoma. Surg Oncol 1996;5:43-8.
- Wittig JC, Bickels J, Kellar-Graney KL, Kim FH, Malawer MM. Osteosarcoma of the proximal humerus: long-term results with limbsparing surgery. Clin Orthop Relat Res 2002;(397):156-76.
- 45. Wittig JC, Bickels J, Kollender Y, Kellar-Graney KL, Meller I, Malawer MM. Palliative forequarter amputation for metastatic carcinoma to the shoulder girdle region: indications, preoperative evaluation, surgical technique, and results. J Surg Oncol 2001;77:105-14.
- Wittig JC, Bickels J, Wodajo F, Kellar-Graney KL, Malawer MM. Constrained total scapula reconstruction after resection of a high-grade sarcoma. Clin Orthop Relat Res 2002;(397):143-55.
- 47. Yang Q, Li J, Yang Z, Li X, Li Z. Limb sparing surgery for bone tumours of the shoulder girdle: the oncological and functional results. Int Orthop 2010;34:869-75. http://dx.doi.org/10.1007/s00264 -009-0857-3