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Limb salvage for malignant bone and soft-tissue tumours of the shoulder girdle

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Abstract We treated 15 patients with malignant bone and soft-tissue tumours of the shoulder girdle using limb salvage operations involving resection of the proximal humerus, scapula and clavicle. The oncological and functional outcomes were evaluated in all patients after an average of 5 (1–13) years. Oncological evaluation revealed that in 11 patients with adequate surgical margins six were continuously disease free and two had no evidence of disease. Three patients with marginal surgical margins had all died. We suggest that only where adequate surgical margins can be obtained should operations be performed. Forequarter amputation is still indicated in patients with complicated neurovascular involvement. Functional evaluation showed that scapular resections, especially glenoid resections, provided poor results (mean score 54%) in comparison with patients without such resection (mean score 82%).

Résumé Nous avons traité 15 malades atteints de tumeurs malignes osseuses ou des parties molles de la ceinture scapulaire en utilisant des opérations du sauvetage du membre qui impliquent une résection de l'humérus proximal, de l'omoplate et de la clavicule. Les résultats oncologiques et fonctionnels ont été évalués pour tous les malades après une moyenne de 5 (1–13) années. L'évaluation oncologique a montré que chez 11 malades avec des marges chirurgicales adéquates six étaient libre de maladie et deux sans évidences de maladie. Trois malades avec une chirurgie marginale sont décédés. Nous pensons que seules les opérations ou une chirurgie à marge adéquate peut être réalisée doivent être faites. L'amputation du membre supérieur est encore indiquée chez les malades avec atteinte neurovasculaire compliquée. L'évaluation fonctionnelle a montré que les résections scapulaires, surtout résections de la glène, don-

naient de mauvais résultats (score moyen 54%) en comparaison de malades sans telle résection (score moyen 82%).

Introduction

The shoulder girdle is the third most common site for malignant bone and soft-tissue tumours. In the past, forequarter amputation had been given as a first choice for treatment for such tumours. The functional result after forequarter amputation was extremely poor, even if better results were achieved oncologically.

Then, since the 1980s, limb salvage has been performed as often as possible. Ideal operations provide a good balance between safe and complete tumour resection and functional reconstruction with no pain after resection. Clinical result of surgical procedures for malignant bone and soft-tissue tumours has developed greatly, with remarkable progress in surgical staging systems, systemic chemotherapy and radiological modalities. However, it is still difficult to indicate the ideal procedure for malignant bone and soft-tissue tumours of the shoulder girdle because of the anatomical complexity and poor existence of surgical barriers.

Between 1988 and 1999, 15 patients with malignant bone and soft-tissue tumours of the shoulder girdle underwent surgery in our department. We retrospectively analysed the adequacy of surgical margins, oncological results, reconstructive techniques and functional results to understanding the relation between the surgical procedure and reconstruction and subsequent functional results.

Patients and methods

The study group comprised 11 men and four women, average age at the operation 45 (9–84) years. Thirteen patients had a primary malignant tumour of the shoulder girdle (four bone sarcomas and nine soft-tissue sarcomas) and two had locally aggressive borderline malignancies (giant-cell tumour of the bone and desmoid

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Table 1 Clinical data. *MFH* malignant fibrous histiocytoma; *MPNST* malignant peripheral nerve sheath tumour; *FVFG* free vascularized fibular bone graft; *Pasteur* pasteurised bone graft;

Lat. dorsi musculocutaneous flap using m. latissimus dorsi; *CDF* continuously disease free; *DOD* dead of disease; *NED* no evidence of disease; *AWD* alive with disease

Case	Age/ gender	Diagnosis (stage)	Resection	Reconstruction	Margin	Follow-up (months)	Functional score (ISOLS)	Status
1	43/M	Giant cell tumour	IIA		Marginal	126	97	CDF
2	47/M	Ewing (IIB)	IIIA		Wide	162	93	DOD
3	9/M	Osteosarcoma (IIB)	VA	FVFG, Lat. dorsi	Wide	80	56	CDF
4	14/F	Osteosarcoma (IIB)	VA	Pasteur, Lat. dorsi	Wide	42	23	CDF
5	71/F	Chondrosarcoma (IIB)	VIA		Wide.	28	80	CDF
6	38/M	MFH (IIB)		Lat. dorsi	Wide	156	80	NED
7	50/M	Liposarcoma (IIB).		Lat. dorsi	Wide	52	93	CDF
8	69/M	Leiomyosarcoma (IIB)			Marginal	46	50	DOD
9	46/M	Synovial sarcoma (IIB)	VIA		Wide	41	20	AWD
10	29/M	Desmoid tumour			Intralesional	40	87	AWD
11	33/M	Synovial sarcoma (IIB)		Lat. dorsi	Wide	39	93	CDF
12	29/M	Angiosarcoma (IIB)			Marginal	26	53	DOD
13	57/F	Myeloma (IIB)		Lat. dorsi	Wide	25	97	CDF
14	73/M	MFH (IIB)			Wide	124	90	NED
15	84/F	MPNST (IIB)	VIA		Wide	28	53	DOD

tumour). All patients with primary bone and soft-tissue tumours underwent systemic and local staging studies; plain roentgenograms of the shoulder girdle and chest, chest CT, bone scan and MRI of the lesion. Pre-operative angiograms were performed in two patients. Open biopsies were performed in all patients. Except one with multiple pulmonary metastases at initial presentation, 14 patients were staged IIB according to the surgical staging system of the American Musculoskeletal Tumor Society.

We used the surgical classification of shoulder-girdle resections from Malawer [8] to classify the surgical resection. The surgical margin was evaluated as radical, wide, marginal or intralesional [1]. Post-operative complications were clarified from the clinical records.

Functional evaluation was carried out according to the ISOLS system by pain, function, emotional acceptance, hand positioning, dexterity and lifting ability [2]. Patients were reviewed every 3 months after surgery. The average follow-up was five (1–13) years.

Results

Clinical presentation

Details of all clinical data is shown in Table 1. Of the five bone tumours, two were osteosarcomas of the proximal humerus. Ten soft-tissue tumours requiring wide resection were synovial sarcoma (2), liposarcoma (2), malignant fibrous histiocytoma (MFH) (2), angiosarcoma (1), malignant peripheral nerve sheath tumour (MPNST) (1), extra-medullary myeloma (1) and desmoid tumour (1). Two patients had had previous operations before referral to our department. Both patients with MFH required additional wide resection and both recurred twice; however, there is no evidence of tumour 12 years after final surgery.

Tumour resection and reconstruction

For the primary bone sarcomas, partial and total scapulectomy (type IIA, type IIIA) and three extra-articular

resections of the proximal humerus, including glenoid and distal end of the clavicle (Tikhoff-Linberg procedure), were performed (type VA). The patient with MPNST had a resection of the chest wall as a part of the wide resection (type VIA).

In no case was vascular reconstruction required. In six patients, we used a latissimus dorsi musculocutaneous flap for primary reconstruction. A massive bone defect after extra-articular resection of the proximal humerus was reconstructed using free vascularised fibular grafts (FVFG) and pasteurised bone. The surgical margins were grossly free of tumour in 14 patients. Microscopic examination revealed, however, marginal margins in two (case 8, 12) and venous invasion in one (case 9).

Adjuvant therapy

Eight patients received systemic chemotherapy. Two patients with osteosarcoma and one with Ewing's sarcoma were treated with neo-adjuvant chemotherapy. One patient with synovial sarcoma required pre-operative intra-arterial chemotherapy (case 9). Others were given post-operative systemic chemotherapy (cases 7, 11, 12, 13). Six patients received radiation therapy after surgery. Two patients with recurrent MFH, and the patient with a huge invasive desmoid tumour, required post-operative radiation therapy (cases 6, 10, 14).

Complications

One deep infection requiring surgical debridement and irrigation occurred in one patient (case 4) (Fig. 1) who underwent extra-articular resection of a proximal humeral osteosarcoma with reconstruction by pasteurised bone. The graft was completely absorbed. The other patient with proximal humeral involvement was reconstructed

Fig. 1 a–b MRI demonstrates a large tumour invading the shoulder joint (T2-weighted image, TR/TE=3000/96) (patient number 4). **c** Plain AP roentgenogram after neo-adjuvant chemotherapy. Tumour volume has decreased. Marginal calcification is seen. **d** Plain roentgenogram 42 months after Tikhoff-Linberg procedure. Marked absorption of the pasteurised humeral bone graft

using a FVFG. Due to a fractured graft, a second grafting procedure was necessary.

Oncological outcome

Of ten patients with wide surgical margins, six were found continuously disease free (CDF) and two had no evidence of disease (NED). Two patients had pulmonary metastases; one was dead of disease (DOD) and one still alive with disease (AWD). Of four patients with inadequate surgical marginal margins, three were dead.

Functional outcome

Functional results are shown in Table 2. Two patients who underwent extra-articular resection of the shoulder joint and total scapulectomy complained of pain after surgery. Six patients had full elevation of the shoulder joint and 13 had full motion and function below the elbow joint. Deficiency below the elbow occurred in two patients with involvement of the radial nerve.

Average functional score was 71% (20–97%). Average functional score of patients who underwent glenoid resection was 54%, whereas it was 82% in patients with no glenoid resection.

Table 2 Functional results according to ISOLS

Case	Pain	Function	Emotional acceptance	Hand positioning	Manual dexterity	Lifting ability	Total	Total (%)
1	5	5	4	5	5	5	29	97
2	5	5	4	5	5	4	28	93
3	5	2	1	2	2	1	13	43
4	3	1	1	1	1	0	7	23
5	5	4	4	4	5	2	24	80
6	5	4	1	5	5	4	24	80
7	5	4	4	5	5	5	28	93
8	3	3	2	3	2	2	15	50
9	3	1	1	1	0	0	6	20
10	5	4	3	4	5	5	26	87
11	5	5	3	5	5	5	28	93
12	3	2	3	3	3	2	16	53
13	5	5	4	5	5	5	29	97
14	5	4	4	5	5	4	27	90
15	5	4	4	4	4	3	24	80

Discussion

Malignant bone and soft-tissue tumours are commonly located around the shoulder girdle. In a review of more than 46,000 cases by the National Cancer Centre, tumours around the shoulder girdle involving scapula, distal clavicle and proximal humerus constituted 12% of all primary malignant bone tumours treated in Japan since 1972. With the great development of radiographic modalities, evaluation of resected specimens and effective systemic chemotherapy, oncological result of limb salvage for malignant bone and soft-tissue tumours has progressed [7]. Functional results after reconstruction following adequate resection, however, are rarely discussed and seem still unsolved.

This study attempted to evaluate retrospectively factors influencing poor oncological results and factors capable of altering post-operative functional results. Our data suggests that resection with inadequate margins induced poor oncological results. All patients with inadequate surgical margins had local recurrence or distant metastasis. In two patients who died of spreading metastasis, we had performed an intentional minimal resection with marginal margins, as the patients strongly opposed a forequarter amputation. Simon has stated that limb salvage must not be inferior to forequarter amputation in regard to prognosis, complications, post-operative function and emotional and social acceptance [15]. Except for one patient, whose venous invasion was first identified histologically after a favourable planned resection with wide surgical margins, we might have performed adequate operation (amputation) for the patients with neurovascular involvement detected by MRI. As were described previously, an inadequate resection makes the patient prone to the risk of local recurrence and subsequent fatal distant metastasis [16].

Limb salvage for tumours of the shoulder girdle was first described by Linberg in 1927. Several authors have since described additional modifications. However, functional results after resection and reconstruction have rarely been discussed. Functional results depend on the

amount of resection. As were previously described [4, 10, 12], results after scapular resection are good when the glenoid is preserved, as also seen in our series. Although glenoid preservation is of great value for good functional results, there are many cases where tumours invade the glenohumeral joint [5]. Since intra-articular resection would result in a high incidence of local recurrence in such cases, it is pertinent to scrutinise the pre-operative radiographic appearance carefully to avoid inadequate operations [6, 11].

The compartmental anatomy of the shoulder and axilla are significantly related to the decision making process for tumour resection around the shoulder [7]. When the brachial plexus and sub-clavian vessels are involved by the tumour, forequarter amputation is indicated. On the other hand, a prospective randomised trial from the National Cancer Institute concluded that the rate of disease-free survival and overall survival of limb salvage versus amputation were comparable [14]. In our series, successful reconstruction using modular prosthesis was done, and flail shoulder using free vascularised fibular graft and pasteured bone graft were performed in two patients only. Among a large number of reports showing good results of bone grafting after removal of the proximal humerus, some authors have shown a relatively high incidence of fractures, infections and local recurrences [3]. In our series, both cases with osteosarcoma of the proximal humerus had complications with the reconstructed bone segment. Marked instability following fracture and deep infection made secondary operations necessary.

Although only 36% of patients who underwent Tikhoff-Linberg procedure with no reconstruction were evaluated as having a good result [7], the average functional result of our patients with performed flail shoulder was moderate (71%), with the exception of two patients with poor results because of radial nerve palsy. Poor results have been reported in patients where a major nerve was sacrificed [13]. Primary reconstructive surgery as tendon transfer to compensate hand function has been proposed [9].

Soft-tissue reconstruction after massive resection is also one of the most important strategies. Gibbons et al. showed the usefulness of the latissimus dorsi musculocutaneous flap to cover preserved tissue [4]. In our series, these reconstructions were evaluated to be of great use and helped avoid skin necrosis induced by tensile skin closure, as well as haematoma formation within the large dead space.

In conclusion, the poor oncological results were caused by inadequate resection and the poor functional results were attributed to following reconstructive procedures. Further efforts must be taken to carefully assess surgical staging and to evaluate individual patients, balancing oncological and functional issues.

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