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ORIGINAL ARTICLE

Segmental Resection and Replantation Have a Role for Selected Advanced Sarcomas in the Upper Limb

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Abstract Although limb salvage surgery for primary sarcoma of the upper limb is a standard procedure, it often is technically challenging. We asked whether segmental resection and replantation would provide (1) local control and long-term survival and (2) useful limb function in patients who had advanced primary malignant and aggressive tumors of the upper limbs. We retrospectively reviewed six patients treated with this procedure when a wide resection around the tumor could not be achieved with other limb-salvage procedures. Diagnoses included osteosarcoma (two), Ewing's sarcoma (one), leiomyosarcoma (one), and giant cell tumor (two). Four patients had displaced pathologic fractures. Minimum followup was 40 months (mean, 164 months; range, 40–214 months). All but one patient remained disease-free; the patient with Ewing's sarcoma died from the disease 40 months after surgery. The average functional score at last followup was 20 points. The mean grasping and pinching power of the operative hand were 66% and 72% of the contralateral side, respectively. Two patients had complications: one had wound dehiscence that subsequently healed and one had radial nerve palsy that recovered spontaneously by 3 months. Segmental resection and replantation may have a role in selected cases for treatment of advanced primary sarcoma or aggressive giant cell tumor of the upper limb as partial limb salvage.

Level of Evidence: Level IV, therapeutic study. See the Guidelines for Authors for a complete description of levels of evidence.

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Each author certifies that his or her institution has approved the human protocol for this investigation, that all investigations were conducted in conformity with ethical principles of research, and that informed consent for participation in the study was obtained.

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Introduction

Various techniques for reconstructing bony and soft tissue defects after tumor resection with wide or radical margins have been described, including the use of autologous vascularized bone graft [14, 19], allograft [8, 9], an endoprosthesis [2, 13, 16, 17, 20], or an allograft-prosthesis composite [3]. However, amputation has been advocated for selective cases of primary sarcoma of the upper extremity when they are large or extensive or when they have neurovascular involvement, local tumor contamination after unplanned surgery, or pathologic fracture [10, 18, 21] of Stage IIB or III [7].

Windhager et al. [22] suggested segmental resection and replantation of the upper extremities as a method of partial limb salvage for upper extremity malignancies that otherwise would be unresectable [5]. The concept of the procedure is similar to that for rotationplasty: segmental resection of the tumor-bearing area of the upper limb as a cylindrical segment, including bone, soft tissue, involved skin, and sometimes vessels and nerves [11, 12, 22]. Although this procedure could retain a relatively functional limb, there are few reports that describe its long-term results [5, 22]. These authors believed resection and replantation could be an alternative to amputation in selected patients with primary sarcoma of the upper limb.

We asked whether segmental resection and replantation would provide (1) local control and long-term survival and (2) useful limb function during long-term followup in patients who had advanced primary malignant and aggressive tumors of the upper limbs.

Materials and Methods

We retrospectively reviewed the records of all six patients who underwent segmental resection and replantation for primary sarcoma or aggressive giant cell tumor (GCT) of the upper limb between 1986 and 1994 (Table 1). The primary indication of the procedure was in Stage IIB primary sarcoma according to the system of Enneking et al. [7] or in Grade III aggressive GCT according to the system by Campanacci et al. [4] when a wide margin around the tumor could not be achieved by other limb salvage procedures because of (1) its size and extensiveness (Patients 3, 5, and 6) or (2) multicompartmental contamination of the tumor cells after unplanned surgery or displaced pathologic fracture (Patients 1, 2, 4, and 5). The diagnoses of all six patients were confirmed by preoperative incisional biopsy and included two osteosarcomas, one Ewing's sarcoma, one leiomyosarcoma, and two GCTs. The site of the tumor was the proximal humerus in three patients, the entire humerus in one, and the forearm in one. Four patients had pathologic fractures at the time of diagnosis: osteosarcoma in one, Ewing's sarcoma in one, and GCT in two. The mean age of the patients (four males, two females) at the time of surgery was 32.2 years (range, 15–42 years). The patients were followed up for a minimum of 40 months (mean, 164 months; range, 40–214 months).

The initial assessment included plain radiographs at the site of the lesion and chest, MRI, computed tomography (CT), arteriography, whole-body bone scan, and chest CT. With these imaging techniques, surgical strategies were planned, including the resection margin of the tumors and neurovascular treatments. In all cases, the neurovascular bundle was free from tumor, although the size and extensiveness of the tumor made limb salvage impossible after wide marginal resection. For preoperative staging, we used the staging system of Enneking et al. [7] for primary sarcoma or the grading system of Campanacci et al. [4] for GCT: four of six were Stage IIB primary sarcomas and the remaining two were Grade III GCTs with no evidence of metastatic lesion (Table 1). We used chemotherapy in four patients; two patients with osteosarcoma received preoperative or postoperative chemotherapy using doxorubicin (80 mg/m^2) and cisplatin (100 mg/m^2); one patient with Ewing's sarcoma was administered doxorubicin (30 mg/m^2), vincristine (1.5 mg/m^2), and cyclophosphamide

Table 1. Clinical data

Patient	Age (years)	Gender	Diagnosis	Site	Stage/grade	Longitudinal tumor length (cm)	Neurovascular involvement	Preoperative CTx/postoperative CTx/RTx	Sacrificed peripheral nerves	Bony link at replantation
1	40	Male	Giant cell tumor	Proximal humerus*	Campanacci Grade III	12.5	No	−/−/−	Axillary	Humerus to distal clavicle
2	26	Male	Osteosarcoma	Proximal humerus*	Enneking Stage IIB	16.3	No	−/+/−	Axillary	Humerus to acromion
3	15	Male	Osteosarcoma	Proximal humerus	Enneking Stage IIB	11.7	No	+/+/−	Axillary	Humerus to acromion
4	32	Male	Giant cell tumor	Proximal humerus*	Campanacci Grade III	8.3	No	−/−/−	Axillary	Humerus to acromion
5	38	Female	Ewing's sarcoma	Entire humerus*	Enneking Stage IIB	23.5	No	−/+/+	Axillary and musculocutaneous	Olecranon to acromion
6	42	Female	Leiomyosarcoma	Distal forearm	Enneking Stage IIB	7.5	No	−/+/−	Superficial and deep radial	Carpal bones to radius and ulna

* These patients had a displaced pathologic fracture at the initial presentation; CTx = chemotherapy; RTx = radiotherapy.

(1200 mg/m^2); and one patient with leiomyosarcoma was administered epirubicin (90 mg/m^2), cyclophosphamide (900 mg/m^2), vincristine (2 mg/m^2), and dacarbazine (300 mg/m^2). In the case of Ewing's sarcoma, radiotherapy was performed as an adjuvant treatment after local recurrence.

The surgical technique used was described previously [1, 5, 22]. Briefly, two circular skin incisions were designed proximal and distal to the region of the tumor, including the biopsy scar, according to the characteristics and site of the tumors. We determined resection margins based on the preoperative MRI, bone scan, and plain radiographs of the lesion. In patients with tumors in the proximal humerus, the patient was placed in the lateral position. After draping, we made the proximal circular incision from the lateral third of the clavicle to the midaxillary line. Longitudinal extension of the incision was made along the course of the brachial plexus and axillary artery leaving the tumor untouched. We divided the pectoralis major muscle just proximal to its distal musculotendinous junction. The axillary sheath was fully exposed after division of the pectoralis minor, the biceps short head, and the coracobrachialis from the coracoid process. With medial traction of the neurovascular bundle at the axillary area, the axillary nerve, posterior circumflex humeral artery, and anterior circumflex humeral artery were ligated and divided. Because all major peripheral nerves were free from any extension of the tumor, the musculocutaneous nerve, radial nerve, and ulnar nerve were traced distally with curative margin to the tumor in all patients with sarcoma by the concept of Kawaguchi et al. [15]. They described the curative surgical margin if the margin is 5 cm or more outside the reactive zone or a surgical margin is outside a barrier with normal tissue between the barrier and the reactive zone of the tumor. This concept has some limitations because their report is from a voluntary group with relatively few controls. However, this barrier concept has been used because of its usefulness in determining the resection margin around the malignant tumors, especially in determining the radial margins. We made the distal circumferential skin incision on the upper arm with sufficient distance from the tumor; we planned the incision to include a curative margin of 5 cm above and below the tumor in patients with primary sarcoma or a wide margin of 2 cm for patients with aggressive GCT. Then, the biceps long head and the brachialis muscles were transected as was the triceps. Proximally, the latissimus dorsi and teres major muscles were transected proximal to their tendinous portions. After the subscapularis muscle was transected at the level of the coracoid process with external rotation of the humerus, the anterior shoulder capsule and the neck of the scapula were exposed. Then we added a posterior skin incision to the anterior incision circumferentially. After the deltoid muscle was transected from the scapular spine and acromion, the supraspinatus, infraspinatus, and teres minor muscles were divided at the level of the coracoid process. The long head of the triceps also was detached to its origin site on the scapula. After soft tissue dissection, the scapula at the neck portion just lateral to the coracoid process was transected, and distal osteotomy followed at the shaft of the humerus with the curative or wide margin to the tumor. In a patient with Ewing's sarcoma involving the entire humerus (Fig. 1), a distal circumferential incision was made around the elbow and the median and ulnar nerves and the brachial artery were dissected free (Fig. 1A). Then we transected the flexor pronator and common extensor muscle groups at the level of the joint. In this patient, the axillary and musculocutaneous nerves were sacrificed proximally, but the other peripheral nerves, including the radial nerve, were dissected and saved because the nerves were tumor-free. In this case, we used the barrier concept by Kawaguchi et al. [15] to achieve a curative margin around the tumor. After removing the tumor-bearing region (Fig. 1B), the axillary artery and its venae commitantes were transected proximally and the brachial artery and its venae commitantes were transected distally to shorten the vessels to prevent redundancy after reconnection of the distal limb. There was no need to resect the median or ulnar nerves because the neurovascular bundle was free from tumors. Then the distal humerus was reconnected to the distal clavicle or acromion using a Gore-Tex® (WL Gore & Associates, Flagstaff, AZ) artificial ligament for static suspension of the replanted limb (Fig. 1C–D). After anastomosis of the major vessels, the major peripheral nerves were placed in loops between muscles (Fig. 1C). Then the triceps was linked to the dorsal scapular muscles and the biceps to the pectoralis major. In the patient with Ewing's sarcoma, the olecranon of the ulna was connected to the acromion with a Gore-Tex® artificial ligament and the flexor-pronator and extensor muscle groups were repaired to the dorsal scapular muscles and the pectoralis major.

In a patient with leiomyosarcoma of the distal forearm (Fig. 2A), we made the proximal circumferential skin incision midforearm. Because the tumor was mainly on the dorsum and the median and ulnar nerves were free from tumor, a longitudinal extension of the incision was made on the anteromedial side of the distal forearm. After exposing the median and ulnar nerves with a curative margin from the tumor according to the concept of Kawaguchi et al. [15], the flexor and extensor muscle and tendon groups were transected proximal and distal to the tumor-bearing region. Then, we made the proximal osteotomy at the midforearm level of the radius and ulna with a curative margin of 5 cm and a distal cut at the level of the radiocarpal and ulnocarpal joint. The radial and ulnar

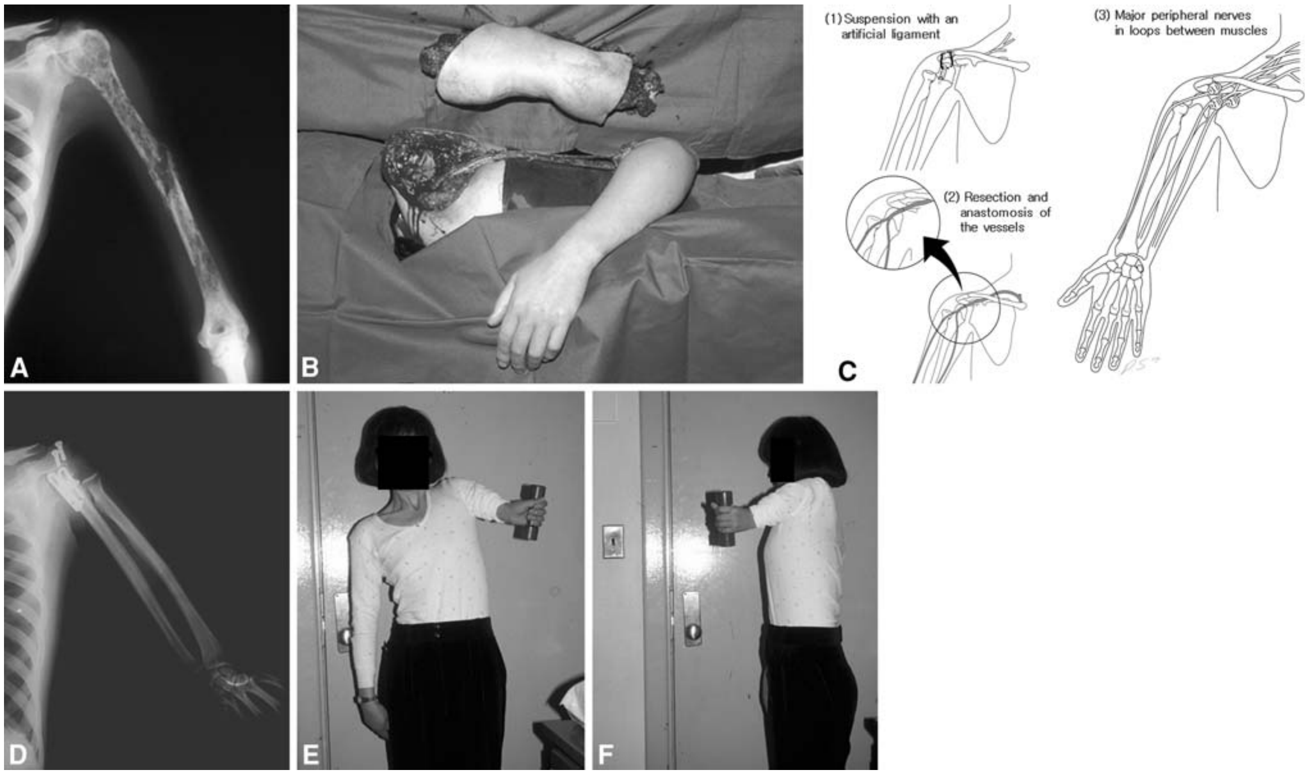

Fig. 1A–F (**A**) A radiograph of a 38-year-old woman with Ewing's sarcoma on the entire left humerus shows a displaced pathologic fracture. (**B**) The tumor and surrounding soft tissue were completely resected while preserving the radial, median, and ulnar nerves. (**C**) After the distal portion was reconnected using a Gore-Tex® artificial ligament between the acromion and olecranon of the ulna, the redundant vessels were removed and anastomosis between the proximal and distal ends was performed. The preserved peripheral nerves then were placed in loops between the muscles. (**D**) The radiograph taken 2 months postoperatively shows the linkage between the acromion and the proximal ulna. She could achieve (**E**) 40° abduction and (**F**) 70° forward flexion, with a score of 19 points according to the functional evaluation system of the Musculoskeletal Tumor Society at 30 months postoperatively. However, she experienced local recurrence 36 months postoperatively and died of pulmonary metastasis 4 months after local recurrence.

arteries and their venae commitantes were exposed and transected proximally and distally (Fig. 2B). We obtained internal fixation between the distal end of the radius and carpal bones with the wrist in 10° extension using multiple Kirschner wires. After the vessels were repaired, the radial and ulnar nerves were placed in loops between muscles. The extensor and flexor tendons and muscles then were repaired in place.

In patients who had replantation in the shoulder region, the replanted limb was immobilized for 1 week with a Velpeau bandage and then another 3 weeks with an arm-neck sling. Active and passive range of motion (ROM) exercise of the fingers was initiated on the first postoperative day. We then gradually instituted active motion of all joints in the limb. In the patient with leiomyosarcoma in the distal forearm, a long arm splint was applied with the elbow at a right angle and the forearm in neutral position to the tips of the fingers and thumb for 2 weeks postoperatively. Then, a short arm cast, below the elbow to just proximal to the metacarpophalangeal joints, was applied for 11 weeks until firm fusion was present on radiographs. In this period, active and passive ROM exercise of the fingers, elbow, and shoulder was encouraged.

Functional evaluation at last followup was performed according to the criteria of the Musculoskeletal Tumor Society [6], which is based on six parameters, including pain, functional activities, emotional acceptance, hand positioning, dexterity, and lifting ability. We also determined grasping and pinching power using a Baseline® hydraulic hand dynamometer (Baseline Corp, Irvington, NY) and rated it as a percentage of the contralateral side. Local recurrences, distant metastases, and complications were recorded.

Results

At the most recent followup, all but one patient was alive and free from any signs of local recurrence or systemic metastasis in the primary sarcoma group; the patients with GCTs also showed no local recurrence (Table 2). One patient with Ewing's sarcoma on the entire humerus

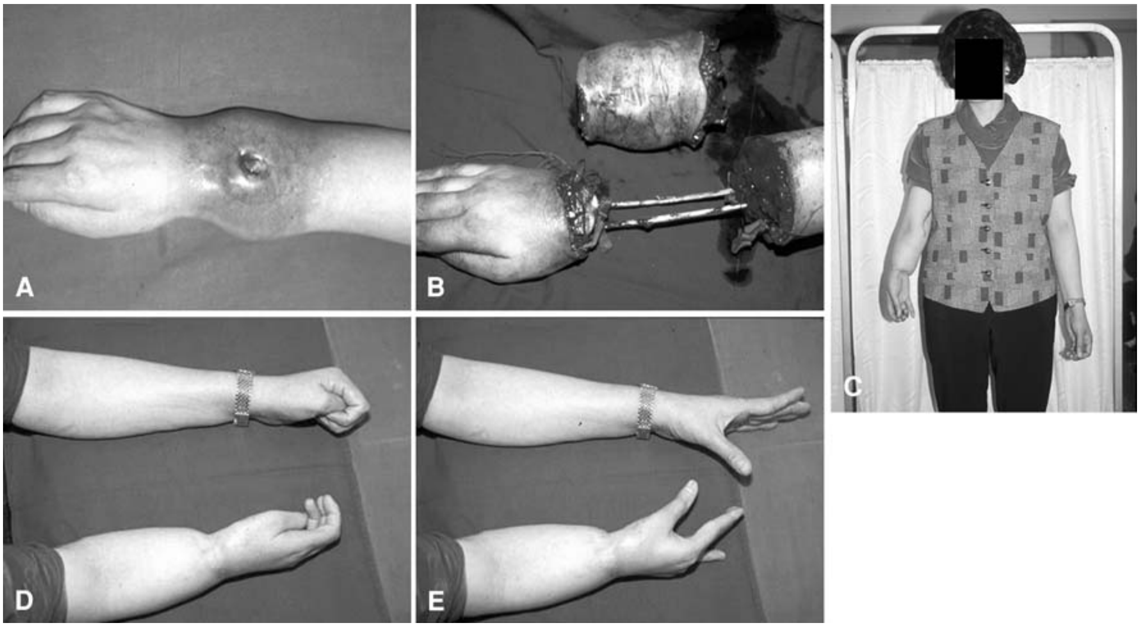

Fig. 2A–E (**A**) A 42-year-old woman with a leiomyosarcoma had a chronically infected skin ulcer of the distal forearm for 3 years. (**B**) The tumor and surrounding soft tissue were completely resected while preserving the median and ulnar nerves. The distal portion then was replanted. At 145 months followup, (**C**) an acceptable appearance, (**D**) useful grip, and (**E**) release of the hand could be achieved, with a score of 24 points according to the functional evaluation system of the Musculoskeletal Tumor Society.

Table 2. Oncologic and functional results

Patient	Followup (months)	Range of motion (°) of shoulder region			Complication	Recurrence/ metastasis	Oncologic outcome	MSTS score	Grasping power (%)/ pinching power (%)
		Abduction	Flexion	Extension					
1	214	50	60	30		−/−	NED	21	67/78
2	210	22	17	20	WD	−/−	NED	13	52/96
3	207	45	40	30	RP	−/−	NED	19	84/91
4	177	55	60	25		−/−	NED	27	90/95
5	40	45	70	35		+/+	DOD	19	75/85
6	145	No value	No value	No value		−/−	NED	24	28/69

Musculoskeletal Tumor Society; WD = wound dehiscence; RP = radial nerve palsy; NED = no evidence of disease; DOD = death of disease.

presented with a pathologic fracture and had local recurrence 36 months postoperatively. Although the patient had forequarter amputation combined with chemotherapy and radiotherapy, she died of pulmonary metastasis 40 months postoperatively.

The average overall functional score was 20 (range, 13–27) for the six patients at last followup according to the functional rating system of the Musculoskeletal Tumor Society. The mean grasping and pinching power of the operative hand was 66% (range, 28%–90%) and 72% (range, 69%–96%) of the contralateral side at last followup, respectively (Table 2). The average ROM of the shoulder in five patients who had the procedure around the shoulder was as follows: abduction 43.4° (range, 22°–55°), flexion 49.4° (range, 17°–70°), and extension 28.0° (range, 20°–35°) (Table 2).

Wound dehiscence occurred in one patient at the site of replantation. The wound healed spontaneously with dressing changes. In another patient, radial nerve palsy occurred and recovered spontaneously 3 months after the operation.

Discussion

Developments in adjuvant therapies, imaging diagnostics, and improved surgical techniques have made limb salvage surgery a standard procedure for most primary sarcomas of the upper limbs. However, amputation rarely would be indicated in selected patients with Enneking Stage IIB or III sarcomas to achieve wide surgical margins around the tumors for local control, which is the primary goal in cancer surgery [5, 22]. In these circumstances, segmental resection of the tumor-bearing region in a cylindrical fashion and replantation of the distal part of the limb with a microsurgical technique might be an alternative procedure to enhance limb function rather than amputation. This segmental resection and replantation procedure uses the same principle as rotationplasty of the lower limb. We determined whether the procedure would provide local control and long-term survival along with useful limb function at long-term followup in patients who had advanced primary malignant and aggressive tumors of the upper limbs.

Limitations of our study include its small sample size and mixed groups that included different tumor and anatomic sites, which meant our results could not be analyzed statistically. Furthermore, this was a retrospective study from one institution. However, there are few patients for whom segmental resection and replantation of the upper limb would be indicated, especially given many are patients who would be treated by amputation to achieve a cure. Additionally, there are few reports describing long-term results of this procedure.

At last followup, all but one patient with sarcoma were alive and free from any signs of local recurrence or systemic metastasis over a mean of 14 years postoperatively. Two previous studies reported only ½ of the patients were alive at the last followup [5, 22]. This might be related to our inclusion criteria for the procedure, which excluded the patients with Stage III sarcoma at diagnosis. One of these studies [22] included patients with Stage III sarcoma who had distant metastasis as an indication of the procedure. Therefore, selection of patients seems to be an important factor in expectation of long-term survival with this procedure.

Our patients' mean grasping and pinching power of the operative hand was 66% and 72% of the contralateral side, respectively (Fig. 2C–E). This grasping and pinching power is probably the result of dissection and sparing of the major peripheral nerves in all our patients. Windhager et al. [22] also suggested unrestricted hand function could be obtained when the radial, median, and ulnar nerves were spared. Consequently, we recommend sparing the major peripheral nerve traveling distally, if possible, to allow superior hand function. In five of our patients who underwent the procedure around the shoulder, functional shoulder motion was maintained as well (Fig. 1E–F), which allowed self-care. We achieved reasonable shoulder stability, perhaps owing to the Gore-Tex® artificial ligament to suspend the distal part of the limb. (We defined stability in this situation as when the patient could voluntarily move the shortened arm [lever arm] based on the artificial ligament used as the fulcrum of the lever arm. The replanted limb should be connected securely to the proximal limb when manual stress is applied to the shoulder region to be considered stable.) The artificial ligament provided restraint for passive motion on axial, longitudinal (compression and elongation of the limb), and translational planes. Our patients' overall functional score was 20 at the last followup according to the functional rating system of the Musculoskeletal Tumor Society, which is comparable to the functional scores of 16 to 23.7 after other limb-salvage procedures such as allografts [9] and endoprostheses [17, 20]. The functional score of the patients probably is correlated with ROM of the shoulder, length of the shortened arm, and power of the pinch and grasp because the total Musculoskeletal Tumor Society score is determined by adding the scores for pain, emotional acceptance, hand positioning, manual dexterity, and lifting ability.

Based on our outcomes, we believe segmental resection and replantation is a reasonable option as a partial limb-salvage procedure in carefully selected patients with Stage IIB primary sarcomas. The procedure can achieve wide surgical margins like those of amputations but maintain useful hand function that prostheses and amputation cannot achieve. However, this technique can be described only as an alternative in extreme situations where all other limb-sparing options are contraindicated.

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