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Concomitant upper extremity soft tissue sarcoma limb-sparing resection and functional reconstruction: assessment of outcomes and costs of surgery

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Abstract
Background The purpose of this study is to investigate functional outcomes and cost impacts of immediate functional reconstruction performed in conjunction with limb-sparing resection of upper extremity soft tissue sarcomas.
Methods Patients undergoing simultaneous limb-sparing upper extremity soft tissue sarcoma resection and functional reconstruction between December 1998 and March 2004 were retrospectively identified, their medical records reviewed, and costs of surgery analyzed. Functional outcomes and patient satisfaction were assessed via patient surveys and the Toronto Extremity Salvage Score (TESS).
Results Thirteen patients met the inclusion criteria. Average follow-up was 43.3 months. Reconstructions included rotational innervated muscle flaps ($n=6$), free innervated myocutaneous flaps ($n=1$), and tendon transfers or grafts ($n=6$). Overall survival was 85 % ($n=11$) and disease-free survival was 77 % ($n=10$). Average total cost of surgery was $26,655. Patients undergoing reconstruction for hand and forearm sarcomas had significantly higher total costs of surgery than those undergoing reconstruction for elbow and upper arm sarcomas. Survey response rate was 91 % ($n=10$). Average TESS score was 76. Of the patients who worked preoperatively, 88 % returned to work postoperatively, and all patients who returned to work currently use their affected limb at work.
Conclusions Patients undergoing immediate functional reconstruction for upper extremity soft tissue sarcoma resection achieved very good to excellent functional outcomes with quick recovery times and a high return-to-work rate following immediate functional reconstruction, thereby minimizing surgical cost impacts. Immediate functional reconstruction in the same surgical setting is thus a viable strategy following upper extremity soft tissue sarcoma resection.

Keywords Cost analysis · Functional reconstruction · Sarcoma · Upper extremity

Introduction

It is estimated that there were 10,980 new cases of soft tissue sarcoma diagnosed in the USA in 2011, with an estimated 3,920 deaths [30]. Approximately 35 and 15 % of soft tissue sarcomas occur in the lower and upper extremities, respectively [36]. Historically, treatment of soft tissue sarcomas with simple excision resulted in very poor outcomes [3] with local recurrence rates estimated at 75–90 % [2, 33].

Treatment of soft tissue sarcomas with limb amputation significantly improved outcomes due to markedly reduced

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local recurrence rates [29, 32], resulting in a 50 % increase in 5-year survival [9]. Limb amputation, therefore, became the standard of care for extremity soft tissue sarcoma despite the morbidity associated with limb loss [14].

In the mid-1970s, wide-margin surgical resection of extremity soft tissue sarcomas with the addition of adjuvant therapy (radiation and/or chemotherapy) achieved recurrence rates, metastasis, and mortality equivalent to that of limb amputation [15, 24, 31, 35]. These advancements in surgical technique, radiation, and adjuvant chemotherapy increased resectability and opportunities for limb reconstruction [25, 27].

Limb-sparing surgery has become the preferred method of treatment for extremity soft tissue sarcomas, with discussion now focusing on the timing, choice, and outcomes of various reconstructive techniques [17, 26, 28]. In cases where adequate tumor resection completely eliminates or compromises crucial limb function, functional reconstructive surgery using innervated muscle or tendon transfers is preferable over non-functional soft tissue reconstruction [1, 8, 11, 12, 17, 22, 28]

We have previously shown that functional reconstructive surgery is costly yet effective in restoring limb utility and improving quality of life parameters following limb-sparing sarcoma resection [23]. However, patient perceptions of functional outcome and surgical cost impacts for immediate upper extremity functional reconstruction have not been directly addressed in prior studies, and comparisons of these parameters between proximal and distal upper extremity functional restoration procedures have not been made.

The purpose of this article is to report functional outcomes and cost comparisons for immediate functional reconstructive surgery of the upper limb following soft tissue sarcoma resection at a high-volume referral center.

Materials and Methods

Patients between December 1998 and March 2004 who underwent functional reconstruction in the same surgical setting as their upper extremity soft tissue sarcoma resection were retrospectively identified in the senior surgeon's database, and their charts were reviewed. The study was approved by our institutional review board. All patients underwent wide tumor resection by the orthopedic oncological surgery team, followed by immediate functional reconstruction by the senior author in the same surgical setting.

Patients were divided into two groups: those undergoing reconstruction for defects proximal to the elbow and those undergoing defects distal to the elbow. The two patient groups were then analyzed along several dimensions: hospital length of stay, intensive care unit days, surgical time, total charges, routine charges, pharmacy charges, radiology charges, laboratory charges, medical supply charges, physical therapy charges, and miscellaneous charges. These data were obtained from internal institutional databases. All monetary charges were adjusted to 2012 US dollars using the United States government's annual Consumer Price Index for All Urban Consumers (CPI-U).

Table 1 Tumor types in the study population

Tumor type	No. of patients (%)
Malignant fibrous histiocytoma	5 (38)
Synovial cell sarcoma	2 (15)
Invasive dedifferentiated osteosarcoma	1 (8)
Clear cell carcinoma	1 (8)
Epithelioid sarcoma	1 (8)
Inflammatory myofibroblastic sarcoma	1 (8)
Other	2 (15)

Functional outcomes and patient satisfaction with reconstruction were assessed via patient surveys and the Toronto Extremity Salvage Score (TESS). This scoring system is a validated longitudinal tool for measuring functional outcomes following resection for soft tissue sarcomas [5–7, 23] and has been shown to be superior for monitoring clinical outcomes as compared to the Musculoskeletal Tumor Society Rating Scales [10] and the Short Form 36 questionnaires [5]. TESS surveys consisted of 30 questions presented as 5-point Likert scales. Questions related to the patient's level of difficulty performing everyday activities. The patient-reported response for each question was summed and then scaled to 100. Lower scores indicate greater difficulty with activities of daily living.

For continuous variables, statistical analysis was performed using unpaired two-tailed t tests. Two-tailed Fisher's exact test was used to compare non-continuous variables. Results were considered significant at an alpha level less than or equal to 0.05.

Results

Thirteen of 49 patients undergoing reconstruction for upper extremity soft tissue sarcoma resection met the inclusion criteria. Average age was 55 years (range 30–87). There were four men and nine women. Average follow-up was

Table 2 Reconstructive methods used for immediate functional restoration following upper extremity soft tissue sarcoma excision

Reconstructive method	No. (%)
Pedicled latissimus dorsi	6 (46)
Tendon allograft	4 (31)
Tendon transfer	2 (15)
Free gracilis	1 (8)

Table 3 Patient information

Patient	Sex	Age	Tumor location	Histology	Tumor grade	Brachytherapy	Radiation	Number of prior excisions	Reconstructive procedure	Complications	Recurrence	Secondary revisions/ procedures	Current disease status	TESS score
1	M	26	Right forearm extensor component	Epithelioid sarcoma	3	No	Preop, postop	2	Non-innervated latissimus free flap; allograft tendons to forearm extensor compartment	None	Yes	Wide local excision of local recurrence; ulnar reconstruction with free fibula flap	Metastatic disease, died of disease	NA
2	F	78	Right biceps	Malignant fibrous histiocytoma	3	No	Postop	5	Innervated rotational latissimus dorsi pedicle flap for biceps function	Intraoperative brachial artery thrombosis	Yes	Radical resection of local recurrence	Metastatic disease, died of disease	NA
3	F	84	Right forearm flexor pronator group	Malignant fibrous histiocytoma	3	No	Preop, postop	0	Tendon transfer of the index, long, and ring finger flexor digitorum superficialis to the flexor carpi radialis	Wrist flexor bulge; wrist hyperextension deformity	No	Tenolysis of flexor tendons	Disease free	61.3
4	F	33	Left humerus and biceps	Dedifferentiated osteosarcoma	3	No	None	2	Innervated gracilis myocutaneous free flap for biceps function	Inferior flap epidermolysis	No	Flap debulking	Disease free	35.3
5	M	29	Right forearm deep flexor compartment	Synovial cell sarcoma	3	Yes	Preop, postop	0	Non-innervated latissimus free flap and tendon transfer (palmaris longus to FPL)	None	No	Radio-ulnar joint arthrodesis with iliac bone graft	Disease free	92.6
6	F	30	Left biceps	Synovial cell sarcoma	3	No	Preop	1	Innervated rotational latissimus dorsi pedicle flap for biceps function	Ulnar nerve paresthesias; chronic donor site pain	No	Ulnar nerve decompression and transposition; flap debulking; forearm flexor muscle lengthening; resection of donor site neuroma	Disease free	98
7	F	75	Left triceps	Malignant fibrous histiocytoma	3	No	Preop	0	Innervated rotational latissimus dorsi pedicle flap for triceps function	None	No	None	Disease free	100
8	M	70	Left biceps and brachioradialis	Undifferentiated sarcoma with myxoid change	3	No	Preop, postop	0	Innervated rotational latissimus dorsi pedicle flap for biceps function	Donor site seroma; incisional cellulitis	No	Resection of benign ring finger and antecubital fossa lesions	Disease free	100

Table 3 (continued)

Patient	Sex	Age	Tumor location	Histology	Tumor grade	Brachytherapy	Radiation	Number of prior excisions	Reconstructive procedure	Complications	Recurrence	Secondary revisions/ procedures	Current disease status	TESS score
9	F	55	Left biceps	Malignant fibrous histiocytoma	3	No	Preop	0	Innervated rotational latissimus dorsi pedicle flap for biceps function	None	No	None	Disease free	NA
10	F	38	Left antecubital fossa and biceps	Dedifferentiated sarcoma	3	No	Preop	0	Innervated rotational latissimus dorsi pedicle flap for biceps function	None	No	None	Disease free	98
11	F	67	Dorsum of left hand	Malignant fibrous histiocytoma	3	Yes	Postop	1	Rotational radial artery forearm flap; delayed allograft tendon to finger extensors	None	No	None	Disease free	56.7
12	M	54	Right wrist	Clear cell sarcoma	3	Yes	Postop	1	Rotational radial artery forearm flap; allograft tendons to extensor compartment	None	No	Extensor tendon tenolysis	Disease free	92
13	F	76	Right radial palm at flexor crease	Inflammatory myofibroblastic sarcoma	2	No	None	0	Non-innervated lateral arm free flap and allograft tendon for index finger flexion	Tendon rupture	Yes	None	Metastatic disease, alive	46.7

43.3 months (range 7–85). Overall survival was 85 % ($n=11$) and disease-free survival was 77 % ($n=10$). 46 % of patients ($n=6$) had undergone prior sarcoma resection. 85 % of patients ($n=11$) achieved local disease control, and 23 % of patients ($n=3$) developed metastatic disease. Of these, 66 % ($n=2$) succumbed to metastatic disease. Excised tumor pathology is presented in Table 1. Average maximum tumor diameter was 5.9 cm (range 1.8–15).

Reconstructive methods and patient parameters are presented in Tables 2 and 3. Seven patients underwent reconstruction proximal to the elbow (Figs. 1, 2, and 3; Video, Supplemental Digital Content 1), and six patients underwent reconstruction distal to the elbow (Figs. 4 and 5; Video, Supplemental Digital Content 2). Of the patients, 23 % of patients ($n=3$) underwent brachytherapy: all had reconstruction for hand and forearm sarcomas; 85 % ($n=11$) underwent radiotherapy; 61 % ($n=8$) and 53 % ($n=7$) received preoperative or postoperative radiotherapy, respectively; and four patients (31 %) received both preoperative and postoperative radiotherapy. Average operative time was 643 min (range 471–925), and average cost of surgery was $26,655 per patient (range $20,924—$40,892).

Complications occurred in six patients (46 %; Table 4). Serious complications, including brachial artery thrombosis, chronic donor site pain, tendon graft rupture, and ulnar nerve paresthesias, occurred in 31 % of patients ($n=6$; Table 3).

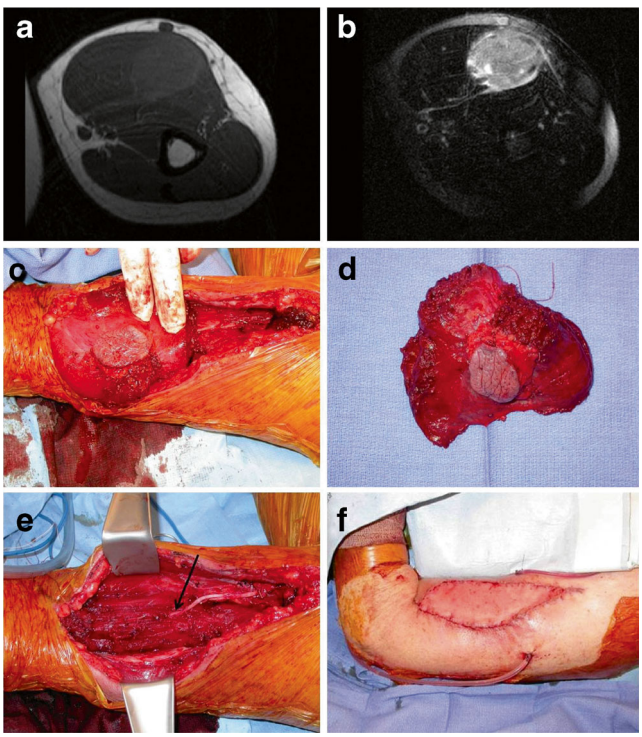

Fig. 1 Proximal upper extremity reconstruction (patient 8). **a** Preoperative T1 and **b** T2-weighted MRI imaging of an undifferentiated sarcoma involving the biceps. **c** Intraoperative view during sarcoma resection (elbow is to the left). **d** Resected specimen. **e** View of postresection defect. Sacrificed musculocutaneous nerve (*arrow*). **f** On-table result following tunneled innervated latissimus flap for restoration of biceps function

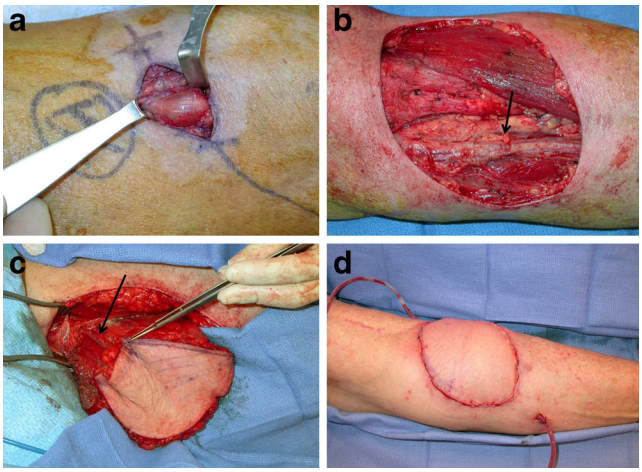

Fig. 2 Patient 8 subsequently developed lesions suspicious for recurrence in the antecubital fossa and ring finger. These proved to be benign. **a** View of antecubital lesion (hand is to the right). **b** Defect following wide local excision. Ulnar artery (*arrow*). **c** A left free gracilis flap was harvested for soft tissue coverage. Flap pedicle (*arrow*). **d** Image of inset flap following end-to-end anastomosis to the ulnar artery

Ten of 11 surviving patients returned the survey in the follow-up period, for an average survey response rate of 91 %. Mean TESS score was 76 (range 36.3 to 100). Average consideration of personal disability was 4.1 out of 5 (1=completely disabled, 2=severely disabled, 3=moderately disabled, 4=mildly disabled, 5=not at all disabled). 88 % of patients (7 of 8) returned to work postoperatively, and all patients who returned to work currently use their affected limb at work. Average time from discharge to return to work was 2.6 months (range 1–4). No patient stated that they would have preferred amputation to limb-sparing surgery with functional reconstruction.

Patients undergoing functional reconstruction for tumor excisions below the elbow had significantly longer hospital stays yet spent significantly less time in the intensive care unit

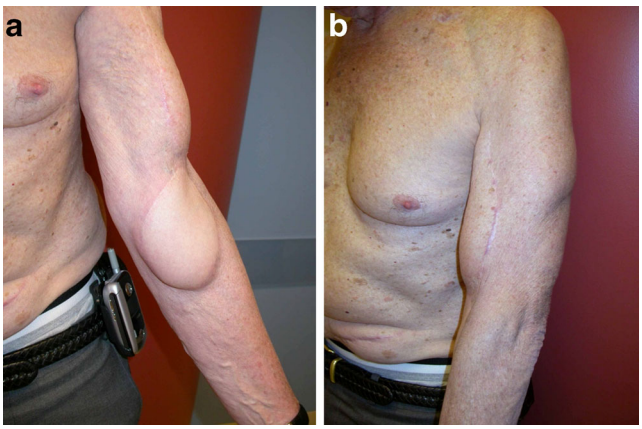

Fig. 3 Patient 8 result. **a** Volar view. **b** Lateral view. Despite the slight bulkiness of the gracilis fasciocutaneous island, the patient was not bothered by this and declined flap debulking

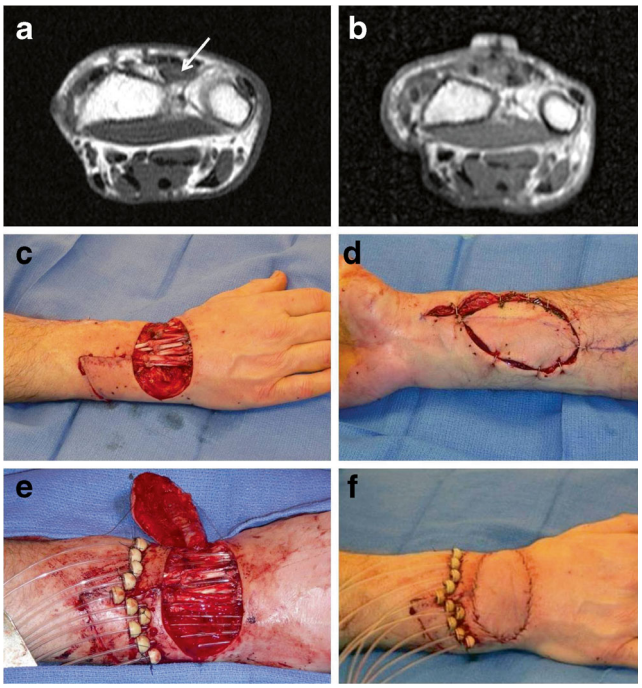

Fig. 4 Distal upper extremity reconstruction (patient 12). **a** Preoperative T1-weighted MRI imaging of a clear cell sarcoma (*arrow*) immediately deep to the hand extensor tendons. **b** T1-weighted MRI 4 months after surgery demonstrating complete sarcoma resection without recurrence. **c** Intraoperative view following clear cell sarcoma resection and reconstruction of extensor tendons with allograft tendons. **d** A pedicled radial artery flap was utilized for soft tissue coverage. **e** Brachytherapy catheters positioned prior to closure. **f** On-table result

than patients undergoing functional reconstruction for tumor excisions below the elbow (8.6 days versus 5.2 days, $p < 0.006$; 0.2 versus 0.7, $p < 0.05$; see Table 5 for full results). Additionally, they had significantly higher total costs of surgery ($31,929 versus $22,764, $p < 0.03$). Furthermore, they received more postoperative radiotherapy (83 % versus 29 %),

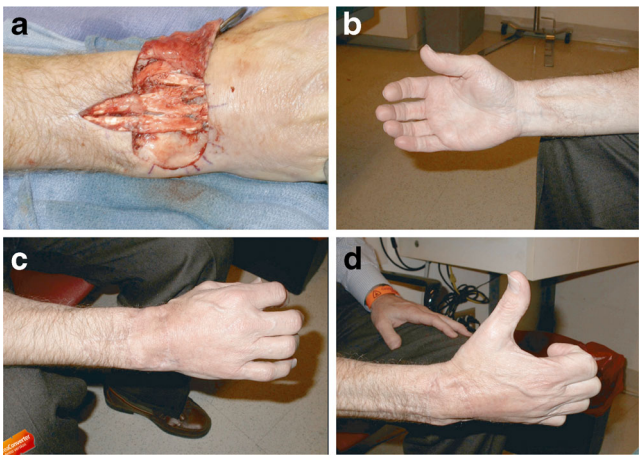

Fig. 5 **a** Patient 12 developed difficulty with wrist extension 9 months postoperatively and underwent tenolysis of allograft tendons. **b** Volar result, **c** dorsal result, and **d** hand with thumb in full abduction 13 months after tenolysis

Table 4 Complications

Complication	Incidence (% of total population $n = 13$)
Incisional cellulitis	2 (15)
Donor site seroma	1 (8)
Chronic donor site pain	1 (8)
Ulnar nerve paresthesias	1 (8)
Partial flap epidermolysis	1 (8)
Intraoperative brachial artery thrombosis	1 (8)
Tendon graft rupture	1 (8)

underwent more surgical revisions (1.0 versus 0.28), and had a higher rate of major complications (50 % versus 20 %) than patients in the proximal reconstruction group, though these differences were not statistically significant (p values 0.07, 0.25, and 0.6, respectively).

Patients undergoing reconstructions proximal to the elbow had higher average TESS scores (86.3 versus 69.9) and a more favorable global consideration of their disability (4.6 versus 3.9), though these comparisons also did not reach statistical significance ($p > 0.10$ in both cases).

Discussion

Although limb-sparing surgery with adjuvant radiation and chemotherapy has become the preferred treatment for upper extremity sarcomas, adequate tumor resection can compromise crucial limb function. Functional reconstructive surgery, with its intuitive benefits in restoring limb utility, should be offered to patients as one of many reconstructive options following sarcoma resection [21].

Despite positive outcomes with functional reconstruction, the adoption of this strategy has not been widespread [26]. Functional muscle is not a prerequisite for limb salvage, and functional reconstruction is more technically demanding, time consuming, and expensive than soft tissue-only reconstruction [13, 16, 19, 23]. Controversy also exists regarding the timing of functional reconstruction, with lack of clarity surrounding the definition of immediate reconstruction [20].

The paucity of data regarding complications, outcomes, and monetary cost of immediate functional reconstruction for upper extremity soft tissue defects following sarcoma resection may explain why this technique is not more widespread. Due to their low incidence, investigators have generally combined upper extremity functional reconstruction with lower extremity functional reconstruction in case series [11, 20, 22, 28]. Conversely, articles addressing upper extremity reconstruction following soft tissue sarcoma resection have had few patients with functional reconstruction [1, 34].

Table 5 Comparison of study parameters by site of resection/reconstruction

	Proximal upper extremity (SD)	Distal upper extremity (SD)	Difference	*p* value
No. of patients	7	6		
Average age	54.1 (20.6)	56 (24.2)	−1.9	>0.1
Average number of prior excisions	1.1 (1.9)	0.7 (0.8)	0.4	>0.1
Average tumor grade	3 (0)	2.8 (0.4)	0.2	>0.1
Average maximum tumor diameter	7.6 (6.4)	4.4 (3.3)	3.2	>0.1
Brachytherapy (%)	0 (0)	50 (55)	−50	>0.1
Radiotherapy (%)	86 (38)	83 (41)	3	>0.1
Preoperative radiotherapy (%)	71 (49)	50 (55)	21	>0.1
Postoperative radiotherapy (%)	29 (49)	83 (41)	−54	>0.1
Length of stay				
Duration (days)	5.2 (1.6)	8.6 (1.9)	−3.4	*0.006*
Intensive care unit (days)	0.7 (0.5)	0.2 (0.4)	0.5	*0.05*
Surgical time (min)	639 (199)	648 (176)	−9	>0.1
Charges (2012 US $)				
Routine	9,638 (2,294)	13,577(4,315)	−3,939	0.08
Operating room	5,834 (1,483)	6,318 (1,029)	−484	>0.1
Pharmacy	1,059 (367)	1,310 (1,896)	−251	>0.1
Radiology	82 (275)	4,124 (7,353)	−4,042	>0.1
Laboratory	1,030 (729)	1,069 (464)	−39	>0.1
Supply	2,495 (700)	2,468 (1,411)	27	>0.1
Therapy	1,194 (872)	1,294 (974)	−100	>0.1
Miscellaneous	1,432 (1,087)	1,769 (558)	−337	>0.1
Total	22,764 (4,188)	31,929 (7,528)	−9,167	*0.03*
Average number of revisional procedures	0.28 (1.2)	1.0 (1.1)	0.72	>0.1
Complication rate (%)	60 (55)	33 (51)	27	>0.1
Major complication rate (%)	20 (45)	50 (54)	−30	>0.1
Percentage completing survey	71	83	−12	>0.1
Average length of follow-up (months)	57.4 (8.6)	40.1 (31.5)	−17.3	>0.1
Average TESS score	86.3 (28.5)	69.9 (21.2)	16.4	>0.1
Consideration of own disability	4.6 (0.9)	3.4 (1.7)	1.2	>0.1
Return to work (%)	75 (5)	100 (0)	−25	>0.1
Time to return to work (months)	2.7 (1.5)	2.5 (1.3)	0.2	>0.1

Significant *p* values in italics

In order to clarify the role of this technique in appropriate patients, our study sought to provide single-surgeon data regarding oncologic, surgical, functional, and monetary cost outcomes of concomitant upper extremity soft tissue sarcoma resection and functional reconstruction in the same surgical setting. As we report a single senior surgeon's experience, the study population is relatively small, limiting its power. This is especially true in light of occasional large standard deviations associated with these data. Patients were not randomized with respect to either functional reconstruction or soft tissue coverage, and consequently, no cost parameter comparisons could be made between these groups. However, single-surgeon studies may control for variability better than larger, multi-institutional studies with respect to technique, thereby providing more consistent outcome data with an associated cost of reduced generalizability. Despite these limitations, patient survey response rate was high, and average follow-up time was adequate.

Patients included in the study had oncologic outcomes consistent with both delayed functional and soft tissue-only reconstructions [1, 20, 22, 28]. Although patient perception of functionality following immediate reconstruction was high, patients undergoing reconstruction for sarcomas distal to the elbow had poorer functional outcomes and considered themselves slightly more disabled than patients undergoing reconstruction for sarcomas proximal to the elbow. They also underwent more surgical revisions and had a higher rate of major complications. While none of these differences were

statistically significant, they are perhaps unsurprising as the distal upper extremity is a much more functionally complex unit than the proximal upper extremity.

The positive outcomes observed in our study were achieved at an average surgical cost of $26,655, surgical time of 643 min, hospital duration of 6.8 days, minor complication rate of 46 %, and major complication rate of 31 %. The significantly increased costs of surgery and lengths of stay in the distal upper extremity group are explained by the higher rates of brachytherapy and postoperative radiotherapy in this group.

Although return-to-work rates and use of limb upon return to work were investigated, only direct inpatient costs were analyzed. No calculations were made regarding the economic impact of patients being out of work or for the costs of revision procedures following functional reconstruction. No adjustments were made for population differences between the proximal and distal reconstruction groups. As tumor excision and functional reconstruction were performed as single-stage procedures, it was not possible to calculate surgical costs and operative times for only the reconstructive portion of each case.

Overall, this study provides more accurate information to address patient concerns regarding immediate functional reconstructive surgery following limb-sparing upper extremity soft tissue sarcoma excision [4]. Additionally, it highlights the need for further cost and outcome investigations for reconstructive procedures in general and for immediate upper extremity functional reconstructive procedures following limb-sparing sarcoma resection in particular.

In summary, following immediate functional reconstruction, patients achieved very good to excellent functional outcomes with quick recovery times and a high return-to-work rate, thus minimizing surgical cost impacts. Functional recovery may be more difficult for patients undergoing resection and immediate reconstruction of sarcomas of the hand/forearm when compared to the elbow/upper arm. Larger studies are needed to evaluate utility differences between various immediate functional reconstructive techniques.

This study supports the notion that concomitant functional restoration should be offered as a reconstructive option in the same surgical setting when oncologic resection will compromise critical upper limb functionality. In this regard, surgeons who possess both detailed knowledge of hand and upper extremity anatomy and can perform functional limb reconstruction through multiple methods appropriate for the patient's functional and tissue coverage needs are a critical component of modern soft tissue sarcoma therapy [34]. Additionally, the presence of such surgeons at the time of tumor resection is crucial to ultimate patient outcomes through the preservation of small distal extremity structures with critical functionality [21]. This is of increasing importance as evidence mounts that smaller tumor-free resection margins, less than 1 cm in recent studies, can achieve long-term disease-free survival in excess of 65 % [18].

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Conflict of Interest The authors declare no conflict of interest.

Statement of Human and Animal Rights All procedures performed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000 and 2008. Informed consent was obtained from all patients for being included in the study.

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