Soft Tissue Sarcomas of the Hand: Functional Reconstruction and Outcome Analysis

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Abstract Functional hand reconstruction following treatment of soft tissue sarcomas (STS) is a difficult surgical problem. Because survival rates between amputation and limb salvage do not differ, there is a trend toward reconstruction. Unlike amputation, hand salvage usually requires multiple complex operations in combination with adjuvant radiation or chemotherapy, prolonged rehabilitation, and carries a high complication rate. We investigated tumor recurrence, survival, and scored functional outcomes to determine if limb salvage is justified after hand STS resection. Patients treated for hand STS between years 1985 and 2005 were reviewed by two surgeons in three medical centers. All patients having functional reconstruction instead of amputation were reviewed. Patient demographics, tumor type and grade, resection extent, reconstruction procedure, timing, adjuvant therapy use, complications, tumor recurrence, survival, and functional outcome were recorded and analyzed. Five patients underwent functional

reconstruction for hand STS. All patients underwent attempted curative resections, and four patients received neoadjuvant or postoperative radiation therapy. Three patients received adjuvant chemotherapy. Reconstructive techniques included three modified pollicizations, one free-tissue transfer, and one groin flap. All patients were alive and disease-free at a mean follow-up of 5 years (range 1.5–17 years). Three patients (60%) had local complications, requiring secondary surgeries. Two complications were related to radiation therapy. Hand function was evaluated using the Enneking Scoring System, and ranged from 17 to 28. The average Enneking score was 22.4, representing an average preservation of 74.6% of function. Because most patients retain excellent function and survival is unaffected, we advocate functional reconstruction despite high complication rates.

Keywords Limb salvage · Hand surgery · Enneking score

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Abbreviations

CMC	carpometacarpal joint
CTX	chemotherapy
ECRL	extensor carpi radialis longus
ECRB	extensor carpi radialis brevis
EDC	extensor digitorum communis
EPB	extensor pollicis brevis
EPL	extensor pollicis longus
FDP	flexor digitorum profundus
FDS	flexor digitorum superficialis
FPL	flexor pollicis longus
MFH	malignant fibrous histiocytoma
MP	metacarpophalangeal joint
PIP	proximal interphalangeal joint
XRT	radiation therapy

Introduction

Soft tissue sarcoma of the hand is a rare diagnosis. In the USA, 3.1 men and women per 100,000 will be diagnosed with some form of soft tissue cancer [22]. Sarcomas account for 1% of adult and 15% of pediatric malignancies. Approximately 15% of all sarcomas occur in the upper extremity, and hand sarcomas comprise less than 3% of all soft tissue sarcomas [10]. Hand sarcomas are more challenging to manage than those in other sites, due to a variety of factors including highly specialized, complex anatomy and a lack of redundant soft tissue [20]. In addition, the combination of an indolent presentation and the overwhelming predominance of benign hand tumors often lead to a lack of urgency in obtaining a diagnosis and a delay in treatment. Although treatment guidelines for soft tissue sarcomas have been developed, a treatment consensus on those tumors involving the hand has not been established because studies have not separated hand tumors from other more common extremity sites [20].

Soft tissue sarcomas of the hand have a higher recurrence rate and worse survival than sarcomas located in other upper extremity sites [4]. As a result, management of these tumors has historically involved radical resection often resulting in amputation because of the proximity of the tumor to vital structures [4]. However, studies showing similar long-term survival with conservative resection and radiation, compared to amputation, have led to a trend toward limb salvage [3, 11, 12, 16]. More than 90% of patients with extremity soft tissue sarcomas undergo limb-preserving surgery currently [17]. Limb-preserving management requires complete resection because patients with positive margins have a 5-year recurrence rate of 16%, despite adjuvant radiation therapy [1, 9]. Combined neoadjuvant chemotherapy and radiation therapy have recently been shown to decrease the size of large high-grade sarcomas, thus potentially increasing the number of candidates for limb salvage [12].

Hand salvage following resection of soft tissue sarcomas is a great challenge for the reconstructive surgeon. Unlike resections in other extremity locations where a wide area of adjacent tissue may be resected with minimal consequence, extirpation of hand sarcomas always compromises vital adjacent structures. While adequate local control via complete resection is mandatory, the endpoints for reconstruction are less clear. Clearly, the goals for reconstruction extend beyond simple limb preservation and must include functional as well as aesthetic considerations [8, 18, 21]. Composite tissue requirements such as skin, tendon, and bone along with the interposition nerve or vascular grafts must be anticipated. If multiple surgical teams are involved, communication is vital so the reconstructive surgeon can anticipate and assess the need for local and free-tissue

transfers. Planning, creativity, and an armamentarium of hand surgery techniques are required.

Because of the extremely low prevalence of soft tissue hand sarcomas, large studies assessing functional outcome after hand salvage do not exist in the literature. In this small series, we investigated the presentation, extent of resection, type of reconstruction, complications, tumor recurrence, patient survival, and long-term functional outcome of patients treated with hand salvage after soft tissue sarcoma resection. The goal of this study was to determine if hand function, tumor recurrence, and patient survival were favorable enough to justify the technically difficult reconstructions, adjuvant treatment, and complications associated with attempted hand salvage.

Materials and Methods

The practices of two surgeons at three institutions between the years 1985 and 2007 were reviewed for patients with soft tissue sarcomas of the hand. Patients who underwent functional reconstruction (hand salvage) following resection were included. A total of five patients with soft tissue sarcomas located distal to the flexion and extension creases of the wrist were identified, and their clinic and hospital charts were reviewed. Following intraoperative confirmation of clear margins by frozen section analysis, a variety of primary reconstructive techniques were employed, chosen on a case-by-case basis by the operating reconstructive surgeon. Postoperatively, patients were started on early aggressive therapy programs that were continued for a minimum of 1 year. The type of tumor, use of adjuvant or neoadjuvant chemoradiation, and extent of resection, complications, tumor recurrence, and functional outcome were documented. Functional outcome was measured using the Enneking functional outcome score, which assesses pain, function, emotional acceptance, hand positioning, manual dexterity, and lifting ability (Table 1). Each of these assessments is scored on a scale of 0 to 5, with 5 as normal anatomic function. Assessments were made a minimum of 1 year following the last surgical procedure. The percent of hand function preserved after surgery is calculated by dividing the patient's Enneking score by the total maximum score of 30 [7]. Patients were then seen at 1 week for a postoperative visit, then again in weeks 3–4. After that, patients were seen at regular follow-up intervals of 7–8 weeks for a minimum of 1 year after surgery.

Results

Five patients underwent curative soft tissue sarcoma resection of the hand with functional reconstruction

Table 1 Enneking scoring system for extremity functional outcome [7, 10].

Score	Pain (P)	Function (F)	Emotional Acceptance (E)	Hand Positioning (H)	Manual Dexterity (M)	Lifting Ability (L)
0	Disabling pain, daily prescription pain medications	Total occupational restriction, completely disabled	Dislike surgery, would not repeat it again	Cannot move arm	Cannot grasp with hand and/or it is completely numb	Cannot even use arm to help other arm
1	Occasional pain, prescription pain medications	Some occupational restrictions, major disability	Accept surgery, would repeat it reluctantly	Cannot lift arm forward above level of waist	Difficulty with basic dexterity such as pinching and/or major loss of sensation	Can only use arm to help other arm in activities
2	Often pain, non-prescription pain medications	Daily occupational restrictions, minor disability	Not fully satisfied with surgery, would still try it again	Have difficulty lifting arm up to level of shoulder	Significant loss of fine movements and/or sensation, inhibited from daily activity	Can only lift arm without any weight
3	Occasional pain, non-prescription pain medication	Occasional restrictions in daily activities, minor disability	Satisfied with surgery, could recommend it to others	Can lift arm up to level of shoulder	Some loss of fine movements and/or sensation, limited daily activity	Can only lift a greatly reduced load than before surgery
4	Occasional discomfort, no pain medications	Occasional restrictions in daily activities, not disabled	Satisfied with surgery, would recommend it to others	Can lift arm over head with some difficulty	Minimal limitations in manual dexterity	Can lift slightly less than before surgery
5	No pain, no pain medications	Not restricted in daily activities, not disabled	Enthusiastic about surgery, would recommend it to others	Can lift arm over head without difficulty	No limitations in manual dexterity (i.e., button shirt, write, etc.)	Can lift as much weight as before surgery

(Table 2). There were three male and two female patients, aged 8 to 46 years old (mean 33.6 years). None of the patients had any comorbidities or other significant medical problems. None of the patients had been diagnosed with cancer previously. Tumor types were: synovial sarcoma ($n=2$), epithelioid sarcoma, rhabdomyosarcoma, and malignant fibrous histiocytoma (MFH). Using the Memorial Sloan–Kettering criteria, three tumors were stage II, and two tumors were stage I [20].

Three modified pollicizations were performed, one of which required microvascular transfer. A second free-tissue transfer, from the contralateral radial forearm, was used to reconstruct a defect after the resection of an epithelioid sarcoma. One patient achieved soft tissue coverage with a pedicled groin flap. Three patients required nerve grafting, secondary to the resection of nerves in close proximity to the sarcoma to achieve clear margins. Four patients received adjuvant therapy. Four patients received radiation therapy (two preoperatively, two postoperatively). Three of the patients receiving radiation also received postoperative chemotherapy. All patients were alive and disease-free at follow-up, which averaged 5 years (range 1.5 to 17 years).

The Enneking Scoring System for extremity functional outcome was used to evaluate patients postoperatively [7]. Postoperative functional scores ranged from 17 to 28 (Table 2). The average Enneking score overall was 22.4, representing preservation of 74.6% of hand function. Despite the challenging reconstructions and postoperative complications, good or excellent hand function scores were achieved in three patients (percent greater than 70). Despite the success of the free-tissue transfer, the patient who developed a significant first web space contracture ulti-

mately achieved a lower functional outcome (Enneking score 17) and fair aesthetic result because of radiation changes to the area (Fig. 1).

Three patients (60%) developed significant postoperative complications, two of which were related to radiation treatment. One patient, who had received neoadjuvant radiation (50 Gy), developed a 2-cm² soft tissue defect over the carpus that presented 3 weeks after reconstruction. This was treated with a local transposition flap. A second patient who received postoperative radiation (64 Gy) suffered radiation burns to the free radial forearm flap used to salvage impending native skin necrosis within the first web space (Fig. 1). There was one nonradiation-related complication: a ruptured tendon graft to the ring finger, which required operative repair (Fig. 2).

Discussion

The rarity of soft tissue sarcomas of the hand has precluded large prospective randomized studies and the development of specific standardized treatment protocols. Surgical resection remains the only potential for cure. Adequate tumor control is paramount, and therefore amputation may be necessary when clear margins cannot be obtained by other surgical approaches [9, 11]. In situations where the pathologist is unsure or the frozen section is unreliable, additional tissue must be sent, or definitive reconstruction should be delayed. The risk of a second anesthetic or suboptimal wound conditions should be subordinate to the risk of a compromised reconstruction that has been contaminated with tumor.

Table 2 Patient demographics and outcomes.

Patient	Diagnosis	Presentation	Resection	Reconstruction	Adjuvant Therapy	Complications	Raw Enneking Score	Percent of Preserved Hand Function
8-year-old girl	MFH (high grade)	Rapid growth hypotenar mass	Skin Digital nerves (ring, small)	Pedicled groin flap Sural nerve grafts	Postoperative XRT Postoperative CTX	None	28	93.3
15-year-old boy	Epithelioid sarcoma (low grade)	Non-painful hypotenar mass	Skin Ulnar artery Digital nerves (ring, small) FDP, FDS (ring, small)	Contralateral radial forearm free flap Palmaris longus grafts Sural nerve grafts	None	Ruptured tendon graft	27	90
26-year-old man	Synovial sarcoma (intermediate grade)	Enlarging non-painful wrist mass	Radial artery ECRL, ECRB, EPL Scaphoid, trapezium, trapezoid Metacarpal (thumb, index)	Transfer MP joint and extensors from index to thumb Neurovascular island flap from index to restore thumb CMC	Preoperative XRT	Skin flap necrosis (requiring transposition flap)	19	63.3
42-year-old man	Rhabdomyosarcoma (high grade)	Non-painful thenar mass	Index metacarpal Subtotal resection thumb metacarpal (CMC preserved) Digital nerves (thumb, index) Digital arteries (thumb, index) FPL Subtotal resection thenar musculature Skin	Transfer index PIP to restore thumb MP Revascularize MP joint with native digital artery and vein graft to radial artery Nerve grafts to thumb Vein grafts to thumb	Postoperative XRT Postoperative CTX	Skin flaps necrosis (requiring radial forearm free flap)	17	56.7
46-year-old woman	Synovial sarcoma (high grade)	Painful wrist mass	Radial artery Trapezium, trapezoid, scaphoid Base of first, second metacarpals EPB	Transfer index MP and EDC to restore thumb CMC Transferred as a vascularized composite graft on ulnar digital neurovascular bundle	Preoperative XRT Postoperative CTX	None	21	70

In treating patients with soft tissue sarcoma of the hand, the reconstructive surgeon must be creative and must have a wide variety of techniques at his disposal if optimal hand function is to be restored. In addition to microsurgical techniques, such as nerve or vessel repair and grafting, a familiarity with a variety of local flaps and bone fixation techniques is necessary. In our series, two patients required a microvascular anastomosis to transfer distant tissue into the defect, and three patients required sural nerve grafts. As with other types of reconstructive surgery, it is important to consider a wide array of disease and patient variables in determining the operative strategy. Case reports from the literature have demonstrated a number of reconstructive

options in managing soft tissue sarcomas of the hand. For example, after resection of a leiomyosarcoma of the hand that included skin, tendon, muscle, and bone, a successful reconstruction was achieved using an iliac crest autograft and dorsalis pedis free flap [14]. Another patient with a fibrohistiocytic sarcoma involving the thumb underwent reconstruction with a sensory free radial forearm flap [2]. Similar to our experience, both of these patients achieved excellent hand function despite complex reconstructive procedures and adjuvant therapy. Although pedicle flaps can be used as in our series, because of the theoretical risks of cross-contamination from the tumor bed, these flaps should be avoided in hand sarcoma patients.

Fig. 1 42-year-old right-hand-dominant man who had undergone a biopsy at an outside institution for an enlarging left thenar mass. The pathology report revealed a high-grade rhabdomyosarcoma. **(a)** The planned incision to include the previous biopsy sight. **(b)** Hash markings reveal the estimated planned osseous resection with preservation of the native thumb CMC joint and distal half of the proximal phalanx. A vascularized transfer of the index finger proximal interphalangeal joint was performed to reconstruct the thumb metacarpophalangeal joint **(c, d)**. After vein and nerve graftings were performed, the skin and soft tissues were redraped, and the incision was closed **(e)**. Mottled areas along the incision progressed to full-thickness skin loss necessitating a free-radial forearm flap to reconstruct the first web space 1 week later **(f)**. Effects of 60-Gy postoperative radiation therapy on the reconstruction 6 months later **(g)**.

The size, location, and involvement of neurovascular structures must be assessed early in the process of establishing the reconstructive plan. Improved preoperative imaging via magnetic resonance imaging has greatly enhanced our ability to make these assessments. When the radial side of the hand is involved, the singular importance of the thumb ray and first web space will dictate reconstruction even at the expense of adjacent digits. Although formal pollicization is certainly an option, in our series, the index finger was used in several ways to reconstitute a subtotally resected thumb. Patient factors including age and occupation are important. Comorbid

disease, functional status, patient expectations, and tolerance for donor site morbidity must be considered.

Integrating the reconstructive plan into the overall treatment strategy is essential to optimize the functional outcome. Communication between extirpative and reconstructive surgical teams, oncologists, and radiation therapists is necessary to determine the type and timing of adjuvant therapy, if indicated. Three of the patients in our review received postoperative chemotherapy. Although adjuvant chemotherapy is controversial, it is generally recommended for rhabdomyosarcomas as well as sarcomas that are high grade or greater than 5 cm [12]. The most

Fig. 2 15-year-old right-hand-dominant boy who presented with a painless palmar mass. A biopsy was performed and revealed a low-grade epithelioid sarcoma. The biopsy site is shown (a). The planned incision incorporates adequate skin margins with an appreciation of the native skin creases (b). The specimen (d) included skin, fat and fascia, a segment of ulnar artery, and the digital nerves and flexor tendons to the ring and small fingers. The resulting defect is shown (c). Palmaris longus tendon grafting from the ipsilateral forearm to the deep flexors of the ring and small fingers was performed (e). After sural nerve grafting (not shown), arterial continuity and soft tissue coverage was achieved via a contralateral radial forearm flow-through flap (f).

commonly used regimens are cyclophosphamide, vincristine, and doxorubicin or etoposide and ifosfamide [12]. While perioperative chemotherapy can certainly debilitate the patient and prolong recovery, the effect on reconstructive outcomes is usually minimal [19]. In contrast, the effect of radiation on functional outcome can be quite significant and must be carefully considered [15].

When combined with resection to negative margins, radiation therapy reduces the recurrence rate for high-grade soft tissue sarcomas of the hand, resulting in survival rates similar to amputation [13]. Because low-grade sarcomas have low recurrence rates, adjuvant radiation is reserved for lesions whose recurrence may result in amputation [13]. Although preoperative radiation does not increase survival, it may decrease tumor size so that limb-sparing resection may be attempted [12, 13].

Despite its beneficial effects on local control, radiation can significantly add to surgical complications [5]. The administration of radiation therapy preoperatively and postoperatively has similar complication rates [6, 15]. Higher complication rates have been noted in small numbers of patients receiving brachytherapy [15]. If preoperative radiation therapy is given, the reconstructive surgeon has the opportunity to assess whether regional radiation-induced changes have occurred and may alter the reconstruction plan to include additional soft tissue coverage if necessary. It should be noted that patients receiving preoperative radiotherapy have frequent wound-healing complications, and in our series, one of the two patients who received preoperative radiation also suffered skin necrosis requiring a local transposition flap. Wound-healing problems after reconstruction involving primary repair, skin graft, or local flap are higher in patients having had preoperative radiotherapy [15]. The combined effects of tension and irradiated, hypovascular soft tissue contribute to this phenomenon and should be anticipated by the reconstructive surgical team. Radiation prior to reconstruction may make dissection more difficult and also produce intimal injury in recipient vessels, an important consideration should revascularization be required.

Postoperative radiation cannot compensate for incomplete resection of primary soft tissue sarcomas of the hand [11]. In addition, postoperative radiation therapy carries a similar complication rate to radiation therapy given preoperatively [15]. However, unlike preoperative radiation therapy, the effect of postoperative radiation cannot be compensated for by the reconstructive surgeon. In addition, the fibrosis and edema caused by radiation therapy is directed at both the transferred tissues (e.g., skin flap, tendon graft) as well as the tumor bed. This is demonstrated in one patient in our series, who was debilitated by an adduction contracture produced by radiation of a radial forearm flap to the first web space. Unfortunately, this

complication was permanent and occurred despite a successful free-tissue transfer. Furthermore, it must be emphasized that early, aggressive occupational therapy is essential to optimize functional outcomes in hand surgery. Because of the effects of postoperative radiation therapy upon the reconstructed hand following tumor resection, this therapy may be delayed or compromised. Because there is no evidence for improved survival or local recurrence outcomes and because complication rates are similar, we recommend preoperative radiation therapy.

Excellent functional outcomes can be achieved in patients with soft tissue sarcoma of the hand, as demonstrated by the average Enneking score of 22.4 in our study. No patient in our series required an amputation as a result of disease recurrence or complication. In addition, all patients remained disease free, and 80% had good or excellent postoperative hand function (defined in our study as functional preservation of 70% or greater). The complication rate is high, especially in patients undergoing radiation treatment. Complications have been shown to result in a decrease in Enneking score [10]. Our complication rate of 60% was similar to other reports [15]. Despite a high complication rate, the resection of vital structures, the use of radiation therapy, and complicated reconstructions, patients can achieve excellent functional outcome. Thus, we advocate that experienced reconstructive surgeons attempt salvage following complete resection of soft tissue sarcomas of the hand, even in the face of complex defects.

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