



Adrienne Anderson. *Parallax View*, 1999-2000. Mixed media on linen, 7' × 6' (feet).

The rarity of sarcomas of the foot and ankle often results in unplanned surgical resection, and further surgery is often required to achieve tumor-free margins.

Unplanned Surgical Excision of Tumors of the Foot and Ankle

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Background: *Soft-tissue tumors of the foot and ankle are relatively common and mostly benign. Thus, many malignant tumors in this region are improperly treated initially. Unplanned excisions can lead to complications that may adversely affect patient outcomes and prognosis.*

Methods: *A retrospective review of patients treated at our institute over a 20-year period for malignant soft-tissue tumors of the foot and ankle was performed. The effect of unplanned surgical excisions on outcomes was examined.*

Results: *When limb salvage was attempted, patients who underwent unplanned surgical excisions had more complications and more extensive surgical procedures involving free flaps, and they were more likely to require adjuvant radiotherapy. No difference in recurrence and disease-free survival was evident between the two patient populations.*

Conclusions: *Despite the lack of statistical power to demonstrate differences in recurrence and survival, unplanned surgical excisions of soft-tissue sarcomas of the foot and ankle probably adversely affect quality of patient care. Suspicious lesions should be referred to surgeons trained in oncologic principles for evaluation and treatment.*

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Introduction

Soft-tissue tumors are common in the foot and ankle. However, it is difficult to differentiate benign from malignant lesions by clinical examination, and thus some malignant soft-tissue tumors are excised inadequately. There are many potential errors of biopsy and treatment of musculoskeletal tumors,^{1,2} and in those patients lacking appropriate diagnostic studies,

unplanned surgical excision is not uncommon.^{3,5} Unplanned surgical excision occurs when tumors are removed without appropriate preoperative evaluation and consideration of the need for obtaining tumor-free margins. Residual disease and tumor recurrence are common in patients who undergo unplanned excision.⁴

We identified a subset of patients who presented with inadequately excised tumors of the foot and ankle that were initially presumed to be benign but, on subsequent pathologic review, were malignant. The reasons for this sequence are related to the rarity of this disease^{6,8} and the variety of specialists who care for patients with foot and ankle problems.³

Although benign tumors in the foot and ankle are relatively common, soft-tissue sarcomas are rare. Approximately 7,800 new cases of soft-tissue sarcoma occur each year,⁶ less than 1% of all adult malignancies.⁹ Less than 10% of these sarcomas occur in the foot and ankle.⁷ In most cases, soft-tissue tumors encountered in the foot and ankle are benign and do not threaten life or limb. Thus, most providers of foot and ankle care remove these tumors with relative impunity and usually with no adverse sequelae.

In some parts of the United States, financial constraints and medical market forces pressure surgeons and nonspecialists to perform procedures for which they lack the appropriate training and support. Even specialists who treat foot and ankle tumors are a diverse group and do not have uniform training in the treatment of musculoskeletal tumors.³ This study analyzes the effects of unplanned surgical excision of tumors of the foot and ankle on treatment and disease outcome.

Table 1. — Diagnosis of 35 Patients With Soft-Tissue Sarcomas*

Diagnosis	Number of Patients	Percent
Synovial sarcoma	12	34.3
Malignant fibrous histiocytoma	9	25.7
Neurofibrosarcoma	4	11.4
Leiomyosarcoma	2	5.7
Liposarcoma	2	5.7
Angiosarcoma	1	2.9
Chondrosarcoma of soft parts	1	2.9
Dermatofibrosarcoma protuberans	1	2.9
Epithelioid sarcoma	1	2.9
Hemangioendothelioma	1	2.9
Sarcoma (NOS)	1	2.9

* Based on a retrospective review of all patients treated at our institute over a 20-year period for soft-tissue tumors of the foot and ankle.

Materials and Methods

All patients within our institutional database who underwent surgery for tumors of the foot and ankle over a 20-year period were reviewed. We identified 43 patients who underwent surgery for a malignant tumor of the foot or ankle. We were interested only in patients with soft-tissue sarcomas and eliminated 5 patients with squamous cell carcinoma, 2 with rhabdomyosarcoma, and 1 with melanoma. Rhabdomyosarcomas were treated on protocols and chemotherapeutic regimens that differed from those used to treat other sarcomas at our institution. A total of 35 patients with soft-tissue sarcomas of the foot or ankle were analyzed.

Clinical records, radiographs, pathology reports, and slides were reviewed retrospectively to determine the incidence and clinical outcome of patients with unplanned surgical resection of sarcomas of the foot and ankle compared to patients with planned excisions. An unplanned surgical excision was defined as a procedure performed for a soft-tissue sarcoma without preoperative imaging or staging studies. Surveillance was maintained in all patients until death. Variables included patient age, sex, location in the foot or ankle, grade, stage, planned vs unplanned excision, type of surgical procedure, complications, local recurrence, metastases, and survival. Tumors were staged using the Musculoskeletal Tumor Society staging system.¹ The student's *t* test and ANOVA were used for statistical analysis. Statistical significance was ascribed at *P* values <.05.

Results

We identified 163 patients who underwent surgery for soft-tissue tumors of the foot and ankle. Thirty-five (21.5%) of these patients had soft-tissue sarcomas. Mean follow-up for this group was 51 months (range = 12-95), and the mean patient age was 44 years (range = 11-96). There were 22 men and 13 women. The most common diagnoses were synovial sarcoma in 12 patients and malignant fibrous histiocytoma in 9 patients (Table 1). Eight tumors (23%) were considered low grade and 27 (77%) were high grade. Most tumors

Table 2. — Tumor Stage of 35 Patients With Soft-Tissue Sarcomas

Stage	Number	Percent
IA	0	0
IB	7	20
IIA	1	2.9
IIB	25	71.4
III	2	5.7

were stage IIB (n = 25, 71.4%); 2 patients presented with stage III metastatic disease (Table 2). The ankle was the location of 13 tumors. Of the 22 tumors in the foot, 4 were in the hindfoot, 11 in the midfoot, 6 in the forefoot, and 1 in the hallux.

Local Recurrence

Overall, 5 (14%) of the 35 patients had a local recurrence. Two of these patients had a planned procedure and 3 had an unplanned procedure followed by attempted wide local resection. The tumors in 4 patients were high grade and 1 was low grade. Only 1 patient with an angiosarcoma of the foot developed metastases and died of disease. This patient had multiple recurrences in the lower extremity requiring higher amputations and eventually developed pulmonary metastases. Tumor margins were histologically positive in only 1 patient following resection. Another patient had a contaminated wide margin in which the mass itself was violated during resection of the tumor. The entire specimen was excised, however, and final margins were negative. Both patients had undergone unplanned surgical resection prior to attempting wide local resection.

Survival

Of the 35 patients, 23 were alive without disease, 2 were alive with disease, 9 died of disease, and 1 died of a myocardial infarction following surgery. The only significant variables affecting survival or tumor-related death were the presence of metastases at the time of presentation or during tumor surveillance and whether the patient had an amputation for local disease control. All deaths due to tumor occurred in patients who had amputations; there were no deaths in the limb salvage group. Age, location, stage, grade, and local recurrence were not significant predictors of survival. Although radiotherapy administration was not significant in predicting survival, chemotherapy was a negative predictor ($P=.05$).

Planned vs Unplanned Resection

Eighteen patients (51%) had an unplanned resection at an outside institution prior to referral, and 17 patients (49%) had their biopsies and index procedures at our institution. Patients with unplanned resections experienced longer delays in receiving definitive treatment from the time of diagnosis (greater than 6 weeks)

Table 3. — Patients With Unplanned Surgical Resections at an Outside Institution Prior to Referral (n = 18)

Patient	Age	Grade	Limb Salvage	Margins	Local Recurrence	Survival	Chemotherapy	Radiation Therapy	Flap
1	15	High	No	Negative	No	NED	Yes	No	NA
2	20	High	No	Negative	No	DOD	No	Yes	NA
3	66	Low	No	Negative	No	NED	No	No	NA
4	26	High	Yes	Negative	No	NED	No	Yes	Yes
5	11	High	Yes	Negative	No	NED	Yes	Yes	No
6	29	High	No	Negative	No	DOD	Yes	Yes	NA
7	25	High	Yes	Negative	No	NED	No	Yes	Yes
8	32	Low	Yes	Negative	Yes	NED	No	Yes	Yes
9	30	High	No	Negative	No	NED	Yes	No	NA
10	86	High	No	Negative	No	NED	No	No	NA
11	55	Low	Yes	Negative	No	NED	No	No	Yes
12	21	High	No	Negative	No	DOD	Yes	Yes	NA
13	61	Low	Yes	Negative	No	NED	No	Yes	No
14	74	High	No	Negative	No	DOD	Yes	No	NA
15	85	High	No	Negative	No	NED	No	Yes	NA
16	29	Low	Yes	Negative	No	NED	No	Yes	No
17	22	High	Yes	Positive	Yes	AWD*	No	No	Yes
18	45	High	Yes	Contaminated	Yes	AWD*	No	Yes	Yes

* These patients are undergoing further treatment.

NED = no evidence of disease

AWD = alive with disease

DOD = dead of disease

NA = not applicable

compared to patients without unplanned resections. They also underwent more complicated surgery involving free flaps when limb salvage was undertaken, had more perioperative complications, and were more likely to be treated with adjuvant radiotherapy. The decision to perform a limb-sparing procedure was determined by tumor depth, size and, most important, involvement of neurovascular structures, deep aponeurotic tissue, and bone.

There were no obvious differences between patients who had unplanned surgical excisions and those who had treatment with regard to local recurrence, limb salvage success, the presence of positive margins, and survival (Table 3).

Discussion

The incidence of soft-tissue sarcoma in our series of patients with soft-tissue tumors of the foot and ankle who underwent surgery at our institution over a period of 20 years was 21.5%. Kirby et al¹¹ reviewed 83 patients with soft-tissue tumors and tumor-like conditions in the foot and ankle and identified 11 patients (13%) with malignant tumors, of which synovial sarcoma was the most common. Chou and Malawer¹² reported 11 malignant tumors in 33 cases of tumors of the foot and ankle treated at a cancer center over a 14-year period. Since the institutions reporting the data were major referral centers, the data do not reflect the true proportion of benign to malignant soft-tissue tumors of the foot and ankle. Foot and ankle sarcomas occur in only 10% of all sarcoma disease sites.⁹ A more accurate estimate of the true incidence of malignant foot and ankle tumors is reported in study by Berlin¹³ in which only 449 sarcomas were diagnosed in an analysis of 307,601 tumors and other lesions of the foot. Most of these lesions were Kaposi's sarcoma, representing 87.1% of this group. Thus, most soft-tissue tumors and tumor-like conditions in the foot and ankle are benign, and treating surgeons rarely suspect a malignancy, especially a sarcoma. This may explain in part the high incidence (51%) of unplanned surgical excision in this group of patients (51%). Comparable results, however, were cited by Davis et al,⁴ who reported 43.5% of patients with soft-tissue sarcoma underwent unplanned resections, and Goodlad and colleagues,⁵ who observed 40% of the patients in their series having unplanned resection. As expected, synovial sarcoma and malignant fibrous histiocytoma were the most common tumors found in the foot and ankle in our series; the majority of tumors (77%) were interpreted as high grade.

In a series of 20 patients with soft-tissue sarcomas of the foot who were treated with combined limb sal-

vage surgery and radiation, Selch et al¹⁴ reported 83% actuarial survival and 63% disease-free survival rates at 3 years. Talbert and associates¹⁵ treated 78 patients with limited surgery and radiation with a mean follow-up of 7.9 years with actuarial 5- and 10-year survival rates of 80% and 69%, respectively. They identified 15 patients with local recurrence but were able to salvage the foot in 12 of these patients. In a retrospective series of 14 patients with synovial sarcoma, Scully et al¹⁶ demonstrated no difference in survival between patients who underwent limb-sparing surgery and those who underwent amputation.

Despite the large number of high-grade tumors ($n = 27$) in our study, only 2 patients presented with metastases and 7 others developed metastatic disease during the course of treatment and follow-up. All of these 9 patients died of disease, all had high-grade tumors, only 1 had a local tumor recurrence, and all had amputations for local disease control. Metastases are less common than expected for high-grade tumors in other, more proximal appendicular sites. In our study, no patient with a low-grade tumor developed metastases. This may lend support for more conservative surgical treatment of these tumors coupled with radiotherapy, which has been espoused by other authors for all sarcomas of the foot and ankle.¹⁴⁻¹⁶

We found a difference in survival in patients who underwent limb salvage vs amputation ($P = .007$), with a larger number of tumor deaths in the amputation group. However, this observation probably reflects a selection bias in that patients with larger, deeper, and more locally aggressive tumors generally underwent amputation, while patients with smaller and more superficial lesions had limb-sparing surgery.

Davis et al⁴ compared outcomes in patients treated primarily in a cancer center vs those treated at non-cancer centers who were referred following unplanned excision. They found that local recurrence was higher in the unplanned surgical excision group, especially patients with residual tumor in the re-resected specimen. Goodlad and associates⁵ reported 95 resections in patients treated in noncancer centers in a series of 236 patients with soft-tissue sarcomas. They found that 59% of those patients with unplanned resections had inadequate margins after re-resection. Clasby and colleagues³ studied 377 patients with primary soft-tissue sarcomas to determine how and by what specialists patients with soft-tissue sarcomas were evaluated and treated. They found that 21.3% of patients were evaluated improperly and only 60% were treated adequately. They noted that outcome was poorest among patients who underwent marginal excision and had tumor recurrence. Paradoxically and contrary to other reported series,



Fig 1. — Clinical photograph of a 21-year-old patient with a synovial sarcoma who underwent an unplanned excision and inadequate follow-up. The tumor recurred and he required definitive treatment.

Lewis et al¹⁷ studied the effects of re-resection in extremity soft-tissue sarcomas and found that 5-year survival was actually better in patients who had undergone prior excision followed by re-excision. They evaluated 685 patients who had primary and definitive resections vs 407 who were believed to have inadequate resection primarily requiring re-resection. Patients treated correctly initially had a mean 5-year survival rate of 70% vs 88% in the re-resection group of patients. This survival advantage was observed controlling for age, tumor grade, depth, size, histology, and margins. The reason for this is unclear; however, the authors conclude that re-resection is indicated in patients with unplanned or inadequate excisions.

In our series, there was no difference in the rate of tumor recurrence or tumor death between patients treated primarily in our institution and those who underwent unplanned surgical excision in another center. Also there was no difference in the rate of tumor recurrence or tumor death. We performed amputations



Fig 2. — Sagittal T1-weighted magnetic resonance image demonstrating a large heterogeneous soft-tissue mass in the plantar aspect of the foot extending from the metatarsal heads to the hindfoot.

immediately on patients with tumors involving bone or effacing or encircling neurovascular structure and on patients with complications resulting from the unplanned surgical excision, specifically hematomas and wound-healing problems. Thus, we selected patients with smaller and more superficial tumors and without complications resulting from the unplanned surgical excision for limb-sparing surgery.

This lack of statistical difference is likely a result of the small number of recurrences and metastasis observed in this study. No patients who had their index or planned surgery at our institution had positive margins at the time of resection. One patient had positive margins and another had wide contaminated margins in the unplanned excision group. The patient with positive margins had a previous unplanned resection through a 10-cm dorsal skin incision for a subcutaneous malignant fibrous histiocytoma. A subsequent magnetic resonance image showed tumor extending 1.5 cm on each side of the incision. A wide local resection was undertaken removing 6 cm of dorsal skin, subcutaneous tissue, and tendon aponeuroses with intra-operative frozen sections margins that were interpreted as negative for tumor. On review of the permanent sections, all margins of resection were focally positive for tumor. The patient with a wide contaminated margin was treated for a mass arising in the sinus tarsi region of his foot. This was excised as an unplanned resection and was found to be a monophasic synovial sarcoma. The patient had preoperative radiotherapy followed by resection. At the time of surgery, the lateral aspect of the calcaneus, the lateral portion of the calcaneocuboid joint, and all of the overlying skin and subcutaneous tissue were removed and a free flap was used to cover the defect. Intraoperatively, as the lateral wall of the calcaneus was reflected, tumor was seen in the plantar muscles. The plantar muscles were then removed widely, and the tumor margins were interpreted as negative on

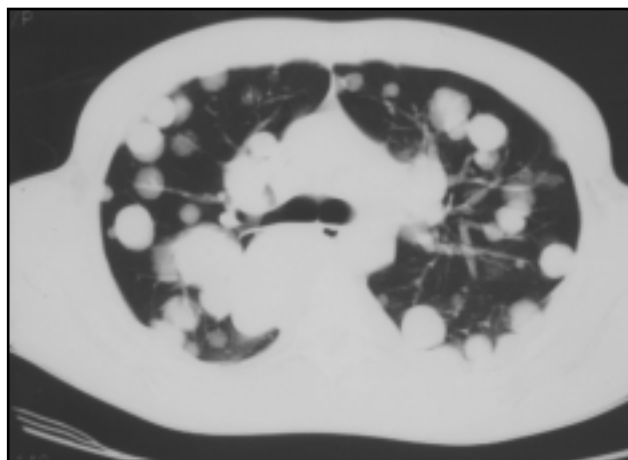


Fig 3. — Computed tomography of the chest with multiple nodules representing metastatic disease.

permanent sections. The tumor recurred proximally in the distal leg requiring amputation. This patient is presently undergoing further treatment.

Seven other patients in this group of unplanned resections had limb-sparing surgery. All were alive and continuously disease-free at latest follow-up. Of 9 patients in the unplanned surgery group who underwent amputation, 5 are disease-free and 4 had distant tumor relapse and died of disease (Figs 1-3).

No differences in local recurrence or survival were seen in patients with planned vs unplanned surgical excisions. However, there were significantly longer delays in treatment in the unplanned surgical excision group. Also, the surgery was more complicated, involving free flaps and skin grafts in 6 of 9 patients who had limb-sparing surgery after unplanned resection compared with none in the patients who underwent limb-sparing surgery as a planned procedure at our institution.

Combined radiotherapy and surgery was used in 7 of 9 patients who had limb-sparing surgery following an unplanned excision and in only 3 of 8 patients without unplanned surgery. There were 3 local recurrences in the unplanned surgery group and 2 recurrences in the planned surgery group. This suggests that when negative margins can be obtained, good oncologic results are possible with surgery alone in some patients. Despite more radical surgery and combined radiotherapy, local recurrence was higher in the unplanned surgery group, although this difference was not statistically significant ($P=.33$). Furthermore, in the 9 patients who had limb salvage after unplanned resection, 6 complications occurred that mostly involved skin slough, flap necrosis, and infection. A superficial wound infection was the only complication in the group of patients with a planned resection.

The use of chemotherapy in nonmetastatic soft-tissue sarcomas is controversial. Chemotherapeutic trials have initially compared single-agent doxorubicin to no treatment and later combined therapy to no treatment.¹⁸⁻²⁰ These trials were conducted in patients with large high-grade tumors because of their significant risk for distant relapse. A potential side benefit in patients who receive preoperative chemotherapy is tumor volume reduction in anticipation of surgery. A meta-analysis was done comparing 13 randomized trials of patients receiving adjuvant chemotherapy for nonmetastatic soft-tissue sarcoma vs cohorts of patients treated without adjuvant chemotherapy.²¹ This analysis demonstrated an overall survival benefit as well as a reduction in local tumor recurrence and distant metastases in chemotherapy-treated patients. Furthermore,

there was a trend favoring multiagent chemotherapy over single-agent therapy.

Only 1 patient received chemotherapy in the limb-sparing group of patients with unplanned surgery, and none received chemotherapy in the planned resection group. As noted, patients who underwent amputation had more aggressive tumors and were more likely to receive chemotherapy. This was observed in 5 of 9 patients in the unplanned surgery group and in only 3 of 9 in the planned resection group. However, there was no difference in local recurrence or survival in patients with amputations in the unplanned vs the planned resection groups. All patients who received chemotherapy had high-grade tumors or advanced disease at the time of presentation. Patients with larger, deeper, and more aggressive tumors, higher-grade lesions, and advanced stage tumors were more likely to receive chemotherapy, and the expected tumor-related outcomes were significantly worse. The role of chemotherapy in patients with nonmetastatic high-grade tumors of the foot and ankle is unclear.

Conclusions

Sarcomas of the foot and ankle are rare and thus are unsuspected in many patients. Unplanned surgical resection is common in these patients and requires further surgery to achieve tumor-free margins.

No difference in local recurrence or disease survival between patients with planned vs unplanned resection was demonstrated in this study. This lack of difference is probably a result of the small sample population and small number of events for these factors. Limb-sparing surgery, however, was technically more difficult, complications were more frequent, and radiotherapy was more commonly used for patients with unplanned surgical excisions. Patients with high-grade, larger, and deeper tumors were more likely to undergo amputation for local disease control. Despite chemotherapy, these patients were more likely to develop metastases and die of their disease. Patients with suspected sarcomas of the foot and ankle should be referred to dedicated cancer centers for definitive treatment.

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