Limb salvage for soft-tissue sarcomas of the foot and ankle

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We evaluated the oncological and functional outcome of 27 patients who had limb salvage for a soft-tissue sarcoma of the foot or ankle between 1992 and 2007, with a mean follow-up of 7.5 years (1.05 to 16.2). There were 12 men and 15 women, with a mean age at presentation of 47 years (12 to 84). Referrals came from other hospitals for 16 patients who had previous biopsy or unplanned excision, and 11 presented de novo. There were 18 tumours located in the foot and nine around the ankle. Synovial sarcoma was the most frequent histological diagnosis. Excision was performed in all cases, with 16 patients requiring plastic surgical reconstruction with 13 free and three local flaps. Adjuvant treatment was undertaken in 20 patients, 18 with radiotherapy and two by chemotherapy. Limb salvage was successful in 26 of the 27 patients. There have been two local recurrences and two mesenchymal metastases. Four patients have died of their sarcoma and two of other causes.

Function was evaluated with the Toronto Extremity Salvage Score and a mean overall score of 89.40 (52.1 to 100) was obtained. A questionnaire revealed that all surviving patients are able to wear normal shoes and none require a walking aid.

Limb salvage can achieve good oncological and functional results with additional treatment.

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Soft-tissue tumours of the foot and ankle are relatively common, but few are malignant. Soft-tissue sarcomas account for 0.63% of all new cases and 1.15% of deaths from cancer in the United States. Fewer than 10% of all soft-tissue sarcomas arise in the foot and ankle.^{1,2} The presence of a sarcoma in the lower limb has been an indication for amputation but in the 1970s and 1980s the combination of conservative surgery and adjuvant radiotherapy made limb salvage more feasible.³⁻⁵

It was not until the 1990s that this approach, combined with plastic surgical reconstruction, was used for soft-tissue sarcomas of the distal part of the limb.⁶⁻¹⁰ Adjuvant radiotherapy can be administered before or after surgery as an external beam or by brachytherapy. External beam techniques are most commonly used. Brachytherapy can be supplemented by external beam radiotherapy, but can also be used as the only form of adjuvant treatment.¹¹ Brachytherapy involves the placement of after-loading catheters into the tumour bed during the operation (Fig. 1). The catheters are loaded in the days after surgery and then eventually removed. This is a very accurate technique, as the bed of the tumour is

under direct vision when the catheters are positioned, and it is generally well tolerated. Catheters can be placed adjacent to free flaps and the periosteum.

Few series have specifically reviewed the management of soft-tissue sarcomas of the foot and ankle.^{7,8,10} The aim of this study was to examine the treatment and functional outcome of soft-tissue sarcomas of the foot and ankle at our regional orthopaedic oncology unit.

Patients and Methods

Between 1992 and 2007, 29 patients presented to our unit with a soft-tissue sarcoma of the foot or ankle. Two patients, seen in 1993 and 1996, had a primary amputation, as at that time preservation of the distal part of the limb was not thought to be possible. Limb salvage was undertaken in 27 patients, and these formed our study group. Amputation was offered as an alternative to the patients who required plastic surgical reconstruction, but none opted for this.

We traced patients from the Queensland Tumour Database and reviewed our medical records. An attempt was made to contact all surviving patients to obtain information from a

Fig. 1

Brachytherapy catheters *in situ* prior to closure of the skin.

questionnaire assessing their current symptoms, occupation, ability to participate in sports/hobbies, footwear, cosmesis and satisfaction with the treatment. Function was evaluated with the Toronto Extremity Salvage Score (TESS).¹² The dates and cause of death of the non-survivors was obtained from the Queensland Cancer Registry at The Cancer Council, Queensland, Australia.

Results

There were 12 men and 15 women, with a mean age at presentation of 47 years (12 to 84). In 16 patients an unplanned excision or biopsy had been carried out prior to referral to our unit; 11 presented primarily. MR scans were performed on all patients, including those who had previous surgery, to image the lesion locally, and a CT scan of the chest was used for distant staging at the time of diagnosis. The mean follow-up was for 7.5 years (1.05 to 16.2) (Table I).

Characteristics of the tumours. Nine were located around the ankle and 18 in the foot. Of those in the foot, seven were on the plantar aspect (Fig. 2), four on the dorsum and three on the medial foot, two each in the hindfoot and big toe. Where the patient presented *de novo*, an open biopsy was performed with an intra-operative frozen section to establish the diagnosis. There were 12 patients with a synovial sarcoma, which was the most common histological subtype (Table I).

Details of treatment. This depended on any previous treatment, the location of the tumour and the histological subtype, and was formulated with close liaison between all members of the multidisciplinary team.

All were fully informed about the options for treatment and the potential complications. Those requiring plastic surgical reconstruction and/or radiotherapy were seen by the appropriate specialist before operation.

All had a wide excision of their tumour or a wider excision of a lesion which had previously been excised inadequately. Plastic surgical reconstruction was required in 16 patients, with 13 having a free flap and three requiring local flaps. Of the free flaps, there were ten from the radial aspect of the forearm (Fig. 3), one from the latissimus dorsi and two from the gracilis (Table II).

One patient had a positive margin after excision. A further excision was performed with an intra-operative frozen section and skin grafting to the bed, which was adjacent to a radial forearm flap. Otherwise all other surgical margins were clear of tumour, although in many cases they were very close.

Adjuvant oncological treatment is summarised in Table I. In general, where a patient presented *de novo*, external beam radiotherapy was performed before operation. If there had been a previous inadequate excision, external beam radiotherapy was carried out after operation, four cases also had brachytherapy. Two patients had chemotherapy as part of their primary treatment. One with a synovial sarcoma had pre-operative chemotherapy, and another with a rhabdomyosarcoma had chemotherapy before and after operation. No adjuvant treatment was used in seven patients.

Post-operative complications (Table II). There was one major wound complication in patient 18 who initially had an intralesional excision of a high-grade myxofibrosarcoma of the lateral aspect of the ankle at another institution. We carried out a wider excision with reconstruction using a radial forearm flap and external beam radiation after operation. An area of the flap failed to heal, became ulcerated, and subsequently resulted in a painful, swollen non-functional limb. After 22 months a below-knee amputation was needed. This was the only failure of a flap, giving an overall survival rate of flaps of 94% (15 of 16) and of the free flaps 92% (12 of 13).

Superficial skin problems occurred in four patients following post-operative radiotherapy. In two who had a free flap, brachytherapy and external beam radiotherapy after operation had delayed wound healing. One patient who had pre-operative radiotherapy developed necrosis of the wound edge which required excision of these areas, and a further patient who had not had any adjunctive treatment had delayed wound healing. A patient aged 72 with multiple medical comorbidities suffered a small post-operative cerebrovascular event.

Oncological outcome. Follow-up data were available for 26 of the 27 patients (Table I). Patient 19 was followed for 13 months after surgery but has since been lost to follow-up, although data from the Queensland Cancer Council indicate that she is still alive. Deaths from the sarcoma occurred in four patients (13.8%). All had pulmonary disease, and two also had extrapulmonary metastases. A further two patients have died of other causes. The mean time from operation to death from the sarcoma was 4.68 years (1.05 to 7.71).

Table I. Histology, adjuvant therapy, current status and Toronto extremity salvage score (TESS)

Patient	Age at presentation (yrs)	Diagnosis	Adjuvant therapy*	Current disease status†	Duration of follow-up (yrs)	TESS
1	44	Synovial sarcoma	Post-operative RT 60 Gy 30 fractions	DFS	3.95	100.00
2	15	Synovial sarcoma	Post-operative RT 50 Gy 25 fractions	DFS	3.57	75.00
3	40	Synovial sarcoma	Brachy 7 catheters 1050 Gy 3 fractions Post-operative 50 Gy 25 fractions	Thigh met (excised)	6.08	52.08
4	21	Synovial sarcoma	None	DFS	8.28	100.00
5	48	Synovial sarcoma	Pre-operative RT 50 Gy 25 fractions	DFS	16.20	100.00
6	12	Synovial sarcoma	Pre-operative RT 50 Gy fractions Post-operative 10 Gy 5 fractions	DFS	9.02	82.26
7	65	Synovial sarcoma	Brachy 7 catheters 800 cGy 4 fractions Post-operative 50 Gy 28 fractions	DFS	12.23	95.54
8	44	Synovial sarcoma	Brachy 7 catheters 1050 cGy 3 fractions Post-operative 50 Gy 25 fractions	DFS	6.04	84.62
9	22	Synovial sarcoma	Post-operative RT 65 Gy 35 fractions	DFS	6.56	-
10	61	Synovial sarcoma	Brachy 7 catheters 1050 cGy 3 fractions Post-operative 50 Gy 25 fractions	DoD (pulmonary disease)	-	-
11	57	Synovial sarcoma	Pre-operative chemotherapy	DoD (pulmonary and extra pulmonary disease)	-	-
12	47	Synovial sarcoma	Brachy 8 catheters 1050 cGy 3 fractions Post-operative 44 Gy 22 fractions	LR, DoD (pulmonary disease)	-	-
13	35	Clear cell sarcoma	Post-operative RT 60 Gy 35 fractions	DFS	1.05	97.58
14	36	Clear cell sarcoma	None	DFS	15.39	-
15	22	Clear cell sarcoma	Brachy 3 catheters 20 Gy Post-operative 50 Gy 27 fractions	DFS	10.73	100.00
16	61	Myxofibrosarcoma	Pre-operative RT 60 Gy 30 fractions	DFS	2.74	90.18
17	84	Myxofibrosarcoma	Post-operative RT 50 Gy 25 fractions	DFS	2.02	71.75
18	60	Myxofibrosarcoma	Post-operative RT 50 Gy 25 fractions, 14 Gy 7 fractions	DFS	6.71	BJA‡
19	62	Malignant fibrous histiocytoma	None	Lost to follow-up	-	-
20	72	Malignant fibrous histiocytoma	Pre-operative RT 60 Gy 30 fractions	D other D	-	-
21	37	Spindle cell liposarcoma	None	DFS	4.42	100.00
22	61	Epithelioid sarcoma	None	DFS	4.65	84.62
23	15	Alveolar rhabdomyosarcoma	Pre- and post-operative chemotherapy	DFS	8.60	100.00
24	54	Acral myxoinflammatory sarcoma	None	LR (excised)	4.14	67.74
25	56	Dermatofibrosarcoma	None	DFS	14.45	100.00
26	60	Pleomorphic sarcoma	Pre-operative RT 50 Gy 25 fractions, 10 Gy 5 fractions	DoD (pulmonary and extra pulmonary disease)	-	-
27	75	Leiomyosarcoma	Post-operative RT 50 Gy 25 fractions	D other D	-	-

* RT, radiotherapy; brachy, brachytherapy

† DFS, disease-free survival; DoD, died of disease; D other D, died of other disease; LR, local recurrence

‡ BKA, below-knee amputation

The Kaplan-Meier analysis for overall survival is shown in Figure 4 and the disease-specific survival in Figure 5.

There have been two local recurrences. This developed in the first (patient 24) 29 months after re-excision of the positive margin for the acral myxoinflammatoryfibroblastic sarcoma. Treatment was by a further excision and a second reconstruction using a radial forearm flap. Histological examination revealed that the recurrence was of a high-grade spindle cell malignant sarcoma and external beam radiation was given, 60 Gy in 30 fractions after operation.

The second local recurrence (patient 12) was of a hind-foot synovial sarcoma, which had initially had an unplanned intralesional excision at the referring centre.

The patient then underwent a wider excision with radial forearm flap and a combination of brachytherapy and external beam radiation. The local recurrence developed after 32 months when the patient returned to the United States, where she was a citizen, to continue her treatment. She ultimately died of the disease.

There have been two ipsilateral limb metastases. Patient 26 developed a lesion in the mid-thigh from a high-grade pleomorphic sarcoma which subsequently also metastasised to the lungs and axilla. The patient has now died of his disease. A further patient (patient 3) developed a recurrence in the groin, which has been excised. In order to achieve a clear margin a vascular resection and soft-tissue reconstruction

Fig. 2

MR scan of a synovial sarcoma on the plantar aspect of the foot.

Fig. 3a

a) Radial forearm flap three years after operation. b) Maximum plantar flexion in the patient in 3a.

Fig. 3b

tion with a transverse rectus abdominis myocutaneous flap was performed. The patient subsequently had external beam radiation to this area, complicated by an infection of the wound which responded to intravenous antibiotics.

Functional scores. We were able to contact 17 of the surviving patients, and so TESS scores were available for only 17 of the 21 survivors. The mean score was 89.4 (52.08 to 100), with the maximum possible score being 100. There was marked variation in the scores. A low score (52.1) was seen in the patient who has recently had a major reconstruction for recurrence in the groin. Low scores were also seen in the patient with a local recurrence who required a second radial forearm flap (52.1), in a woman aged 86 with degenerative disease in several other joints (71.1) and in a man who had previously had surgery on his ipsilateral hip

and ankle following a road traffic accident (84.6). Of the four patients whose scores were assessed, one had required a below-knee amputation for failed limb salvage, and one has been lost to follow-up. The other two could not be contacted at the time of the study, but both were functioning well at their last follow-up.

The mean TESS score for the patients who had plastic surgical reconstruction was 85.6 (52.1 to 100), and for those without, 96.3 (82.3 to 100). This was not significant using the Mann-Whitney U test.

The mean TESS score for patients who presented to our unit *de novo* was 91.4 (82.3 to 100) and 87.0 (52.1 to 100) for those who had prior surgery at another institution. This was not significant using the Mann Whitney U test.

Questionnaires. All 17 patients who were contacted completed a questionnaire. All the patients were wearing normal shoes, and only one required an orthosis. None used a walking aid. Of the patients in employment, all except the man who had recent surgery for a metastasis in the groin, had returned to their pre-operative occupation. There were two patients who were unhappy with the appearance of their foot/ankle, one was accepting and the remainder were happy.

No pain was experienced in 11 patients, four had occasional pain, and two described constant discomfort, but only one took simple analgesics.

Eight patients described swelling, particularly in hot weather. Of the eight patients who like to participate in sports, six are still able to continue. A further two patients were not able to walk barefoot on the beach or surf since their surgery.

All patients thought that their outcome was preferable to having a below-knee amputation. All were satisfied with their treatment overall, and only one was unsure whether he would have the same management.

Discussion

Soft-tissue sarcomas of the foot and ankle present a challenge to achieve limb salvage with acceptable functional and oncologic outcomes. Despite what may seem in some cases to be a lengthy programme of treatment, with the potential for complications and a poor outcome, we have shown that good results can be achieved, with preservation of the limb by a combined approach. We had only one failure of treatment, requiring amputation, and this compares well with other published series, which have lower rates of reconstruction by plastic surgery. Colterjohn et al⁸ also had one failure in 30 patients (97%) and Talbert et al,⁶ Jyothirmayi et al⁹ and Selch et al⁷ described similar results.

Plastic surgical reconstruction was performed in 60% of our patients, the majority having radial forearm fasciocutaneous flaps. In a review of 25 cases of foot and ankle reconstruction using this flap, Weinzwieg and Davies¹³ had a success rate of 92% when used for diverse indications, including three tumours. They concluded that this flap meets most of the anatomical requirements for the ideal foot flap. It

Table II. Surgical details and complications

Patient	Location	NV* structure sacrificed	Previous biopsy or unplanned excision	Plastic surgery	Flap complications	Other complications†
1	Foot - plantar	All contents 1st & 2nd interspaces	Yes	Radial forearm flap	None	
2	Foot - dorsum	All dorsal NV structures	Yes	Radial forearm flap	None	
3	Ankle - medial	None	Yes	Plantar vascularised	Slow-healing ulcer	
4	Foot - great toe	NV structures to 3rd toe	Yes	None	N/A†	
5	Foot - plantar	None	Yes	Gracilis flap	None	
6	Ankle - medial	Dorsalis pedis and deep peroneal nerve		None	N/A	Wound edge necrosis, excision of wound edges
7	Ankle - lateral	Posterior tibial nerve and artery	Yes	None	N/A	Radiation burn
8	Foot - plantar	Medial NV bundle to great toe	Yes	None	N/A	Superficial blistering post RT
9	Ankle - medial	None	Yes	None	N/A	
10	Foot - medial	None		None	N/A	
11	Foot - medial	All NV structures to 1st and 2nd rays		None	N/A	
12	Foot - hindfoot	Posterior tibial artery	Yes	Radial forearm flap	Flap infection, ulcer, declined amputation	
13	Foot - dorsum	Dorsalis pedis artery	Yes	Radial forearm flap	None	
14	Foot - plantar	Digital nerves to index and middle toes		None	N/A	
15	Foot - plantar	Posterior tibial nerve and artery		Local rotation flap	None	
16	Ankle - lateral	Sural, deep and superficial peroneal nerves		Gracilis flap	None	
17	Foot - dorsum	None	Yes	Radial forearm flap	None	
18	Ankle - lateral	None	Yes	Radial forearm flap	Persistent non-healing ulcer	CRPS, painful non-functioning limb. BKA
19	Foot - medial	None	Yes	Dorsalis pedis flap	Delayed wound healing	
20	Ankle - lateral	None		Latissimus dorsi flap	None	Small CVA post-operative, multiple medical problems
21	Foot - plantar	None	Yes	None	N/A	
22	Foot - hindfoot	None		Radial forearm flap	None	
23	Foot - plantar	NV structures to 5th ray		Radial forearm flap	None	
24	Foot - dorsum	None	Yes	Radial forearm flap	None	Infective radiation dermatitis
25	Foot - great toe	Digital nerves and arteries to great toe	Yes	None	N/A	
26	Ankle - medial	None		Radial forearm flap	None	
27	Ankle - medial	Posterior tibial nerve and artery		None	N/A	Local inflammation with patchy skin loss post RT

* NV, neurovascular

† NA, not available

‡ RT, radiotherapy; CRPS, complex regional pain syndrome; BKA, below-knee amputation; CVA, cerebrovascular accident

facilitates the restoration of normal foot contour by replacing 'like with like', allowing patients to use normal shoes without the need for debulking, and it provides a durable and stable weight-bearing plantar surface when walking. All of our patients were able to return to wearing normal shoes.

Sarcomas of the distal extremity have a better survival than sarcomas located elsewhere. Zeytoonjian et al¹ showed an overall death rate from all sarcomas of 26.6%, and from the foot and ankle of 10.3%. That in our series was 14.8%. The reasons for the improved survival include the fact that small lesions in the foot and ankle are often noticed early because they interfere with function. A tumour which is small, noticed early, and in a distal part of the limb, which has a relatively limited blood supply, is less

likely to metastasise and cause death.¹ Given the improved survival of sarcomas in this location, our treatment regimen would seem justified.

An unplanned excision or biopsy had been carried out on 16 patients (56%) prior to referral to our unit. This is a relatively common occurrence in soft-tissue sarcomas of the foot and ankle, given that benign lesions outnumber malignant ones by 100 to 1.¹⁴ Temple et al² observed that 51% of their foot and ankle tumours had unplanned excisions at other institutions. These patients needed more complicated surgery, had more peri-operative complications, and were more likely to be treated with adjuvant radiotherapy. However, there were no differences in the oncological outcomes.

Fig. 4

Kaplan-Meier survival showing overall survival.

Fig. 5

Kaplan-Meier survival showing disease-specific survival.

The TESS has demonstrated reliability, validity and responsiveness in injury to the lower limb.¹² The mean overall score in our patients was 89.4 (52.1 to 100), which is comparable with other published series for this location of 88.1.¹⁵ The questionnaire responses demonstrate a high rate of satisfaction. We have not performed a primary below-knee amputation in the last 12 years. The benefits of time-consuming surgical reconstruction and adjuvant therapies are often questioned, given the excellent functional

results that can be obtained with below-knee amputation. Comparisons of the quality of life between limb salvage and amputation for sarcomas of the lower extremity show little difference,^{16,17} but these studies were not specific for the distal part. The presence of a soft-tissue sarcoma in the foot or ankle is not an indication for amputation. Good functional and oncological results can be achieved with a combination of excision of the tumour and, where required, plastic surgical reconstruction and/or adjuvant therapies.

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