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Limb salvage in aggressive and malignant tumours of the fibula

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Abstract We treated 25 patients with aggressive and malignant fibular tumours between April 1989 and May 2001. There were 11 osteosarcomas, seven Ewing's sarcoma and five chondrosarcomas. The tumours predominantly involved the upper one third; 16 were of Stage IIB. Neo-adjuvant and adjuvant chemotherapy were given to all patients with osteosarcoma and Ewing's sarcoma. In 20 patients, surgical margins were wide and in three radical. Mean follow-up was 71 (26–168) months. Local recurrences were met with in three patients requiring amputation. Three patients died of disease and one was lost to follow-up. Functional outcome was excellent in 12 patients and good in seven. The 5- and 10-year survival rates of patient and limb are reported based on Kaplan-Meier survival analysis.

Résumé Entre avril 1989 et mai 2001 nous avons traité 25 malades avec une tumeur fibulaire maligne et agressive. Il y avait 11 ostéosarcomes, sept sarcomes de Ewing et cinq chondrosarcomes. La majorité des tumeurs concernait le tiers supérieur. Seize étaient de stade II B. Tous les malades avec un ostéosarcome ou un sarcome d'Ewing ont reçus une chimiothérapie néoadjuvante et adjuvante. Pour 20 malades les marges chirurgicales étaient larges et pour trois radicales. La moyenne de suivi était de 71 mois (26–168). Une récidive locale est survenue chez trois patients, exigeant l'amputation. Trois malades sont morts de maladie et un a été perdu de vue. Le résultat fonctionnel était excellent pour 12 et bon pour sept malades. La survie à 5 et 10 ans estimée pour les malade et pour le membre est rapportée selon l'analyse de Kaplan-Meier.

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Introduction

Fibular resections have been contemplated as a viable option for salvaging limbs without skeletal reconstruction, improving the oncological outcome and survival of patients undergoing such procedures for aggressive and malignant fibular tumours [8, 12]. Accurate staging of the tumour [11], authoritative pre-operative histopathological diagnosis and adequate oncological margins are mandatory pre-requisites for achieving the desired outcome in fibulectomies.

Quite a few techniques of fibular resections have been described in the literature [8–10]. Resection of tumours at various sites of the fibula presents problems of oncological adequacy, ankle instability and, to a lesser extent, instability of the knee [1, 3, 6]. Early pre-disposition for spread to the adjacent peroneal nerve and muscles is a major determinant in achieving oncologically satisfactory resection margins. In our present series of aggressive and malignant tumours of various sites of the fibula, we have evaluated oncological outcome, functional results and 5- and 10-year survival rates of the patient and the limb following partial and total fibulectomies.

Materials and methods

Twenty-five patients with aggressive and malignant fibular tumours were surgically managed with en bloc resection between April 1989 and May 2001. Of them, 18 were males and seven were females ranging from 10 to 55

(mean 18.4) years. The lesions predominantly involved the upper third in 17 patients and the middle third in six (Table 1). While nine patients were referred to us after an open biopsy, we reached a pre-operative diagnosis by closed needle biopsy in the rest.

Staging evaluation was done with the help of plain radiographs in all patients, CT scan in 14, MRI in five, technetium isotope bone scan in nine and angiography in three. The majority presented as Stage IIB of Enneking's system. Osteosarcoma dominated the series with 11 patients, followed by seven with Ewing's sarcoma. Histopathological diagnoses and the staging are given in Table 1.

Wide margins of resection were achieved in 20 patients. Three patients required radical resection of the entire compartment. A patient with an aneurismal bone cyst and another with an aggressive giant cell tumour, both involving the proximal fibulas, were given a marginal clearance (Table 1). The three patients who required radical resections had aggressive extra-compartmental osteosarcoma and Ewing's sarcoma involving the peroneal nerve and the anterior tibial artery. Such radical resections were almost similar to that of Malawer type II fibular resections [8], i.e. removal of the anterior and lateral muscular compartments, peroneal nerve and anterior tibial artery with an extra-articular resection of the superior tibiofibular joint. All those who underwent proximal fibular resections had a concomitant repair of the fibular collateral ligament. No reconstruction was employed for distal fibular resections [10]. A lateral gastrocnemius flap was used to cover the resected defect. Neo-adjuvant and adjuvant chemotherapy were given to all patients with osteosarcoma and Ewing's sarcoma depending on the regimen prevalent at the time of presentation. During the initial years, pre-operative radiotherapy was given to two patients with Ewing's sarcoma.

Post-operatively, the limbs of those who underwent distal fibulectomy were immobilised with a short leg cast for a minimum period of 3 weeks to allow capsular healing and soft-tissue fibrosis. Other patients were encouraged to undertake early weight bearing as and when it was tolerable, supported by and ankle-foot orthosis in those who underwent radical resections. All patients were followed up monthly for 6 months and 6-monthly thereafter with clinical examination and plain radiography.

Table 1 Twenty-five patients with malignant and aggressive fibular tumours

No.	Age	Gender	Diagnosis	Site	Stage	Pre-op. chemotherapy	Margins	Post-op. chemotherapy	Follow-up in months	Functional outcome	Oncological outcome
1	20	M	Ewing's	U-1/3	II B	Yes	Wide	Yes	147	Good	DOD
2	10	F	Ewing's	M-1/3	II B	Yes	Radical	Yes	169	Fair	CDF
3	14	M	Ewing's	U-1/3	II B	Yes	Wide	Yes	157	Excellent	NED
4	17	M	Chondrosarcoma	U-1/3	I B	No	Wide	No	117	Excellent	CDF
5	13	M	Ewing's	M-1/3	II B	Yes	Radical	Yes	54	Good	DOD
6	18	M	Chondrosarcoma	U-1/3	II A	No	Wide	No	101	Excellent	CDF
7	19	M	Osteosarcoma	U-1/3	II B	Yes	Wide	Yes	99	Good	NED
8	20	F	Osteosarcoma	U-1/3	II B	Yes	Wide	Yes	56	Good	DOD
9	17	F	Osteosarcoma	U-1/3	II B	Yes	Radical	Yes	73	Fair	CDF
10	20	M	Osteosarcoma	U-1/3	II B	Yes	Wide	Yes	69	Excellent	CDF
11	15	M	Osteosarcoma	U-1/3	II B	Yes	Wide	Yes	67	Fair	CDF
12	12	F	Osteosarcoma	U-1/3	I B	Yes	Wide	Yes	63	Excellent	CDF
13	16	M	Osteosarcoma	U-1/3	II B	Yes	Wide	Yes	54	Fair	CDF
14	55	M	Chondrosarcoma	M-1/3	III B	No	Wide	No	44	Good	DOC
15	17	M	Osteosarcoma	U-1/3	II B	Yes	Wide	Yes	41	Excellent	CDF
16	16	M	Ewing's	M-1/3	II B	Yes	Wide	Yes	40	Excellent	CDF
17	25	M	Chondrosarcoma	D-1/3	I B	No	Wide	No	36	Good	CDF
18	14	M	Osteosarcoma	U-1/3	II B	Yes	Wide	Yes	33	Good	CDF
19	18	M	Chondrosarcoma	M-1/3	I B	No	Wide	No	98	Excellent	CDF
20	27	M	Osteosarcoma	U-1/3	I B	Yes	Wide	Yes	48	Excellent	CDF
21	10	F	Osteosarcoma	U-1/3	II B	Yes	Wide	Yes	76	Poor	NED
22	13	F	Ewing's	D-1/3	II B	Yes	Wide	Yes	26	Poor	CDF
23	13	M	Ewing's	M-1/3	II B	Yes	Wide	Yes	28	Excellent	CDF
24	31	M	GCT	U-1/3	II A	No	Marginal	No	60	Excellent	CDF
25	12	F	ABC	U-1/3	II A	No	Marginal	No	28	Excellent	CDF

GCT, giant cell tumour; ABC, aneurysmal bone cyst; CDF, continuously disease free; NED, no evidence of disease; DOD, died of disease; DOC, died of other causes.

Results

The average period of follow-up was 71 (26–168) months. One patient with an osteosarcoma was lost to follow-up after 54 months. Post-operatively, two patients had superficial wound infections and one developed flap necrosis requiring skin cover. Two patients with osteosarcoma and one with Ewing's sarcoma developed local recurrence that necessitated above-knee amputations. Two patients who underwent wide resection of the proximal one third of the fibula developed peroneal nerve palsy, of which one had a partial recovery within 6 months.

Functional outcome was assessed using Enneking's functional evaluation system [4, 5], which includes motion, pain, stability, deformity, strength, functional activity and emotional acceptance as measuring criteria. Thirteen patients had an excellent outcome, six good, three fair and two poor at their latest followup (Table 1).

The various histopathological diagnoses with their oncological outcomes are also given in Table 1. Seventeen patients were continuously disease-free, and three patients had no evidence of disease after limb ablation for local recurrence. Three other patients died of disease following widespread metastatic lesions. One patient with chondrosarcoma, who was continuously disease-free, died of unrelated renal disease. The 5-year survival rates were derived using the Kaplan-Meier survivorship method. Death was taken as the end point for patient survival and amputation for survival of the limb. The 5- and 10-year survival of the patient was 89% and 74%, respectively, and survival of the limb was 54% at 5 and 10 years, as depicted in Fig. 1.

Discussion

The fibula is a rare site (2.5%) for primary bone tumours [2]. Few reports are available regarding limb-salvage techniques after resection and their eventual outcomes. The fibula also presents certain anatomic peculiarities. The thin cortices together with multiple muscular attachments make the lesions become extra-compartmental much earlier than in other sites. Hence, prompt surgical intervention can give adequate oncological margins. We also agree with Malawer [8] who described the high frequency of direct muscle invasion by tumours of the proximal fibula in contrast to the pushing borders usually observed with tumours in other anatomical locations. This fact also emphasises the role of effective neo-adjuvant chemotherapy to achieve tumoricidal evidences well away from the margins of resection. This in turn serves as a good predictor of the efficacy of the neo-adjuvant regimen that was instituted, which helps to plan the post-operative adjuvant therapy [7]. Our series of fibular resections has shown a local recurrence rate of 12%, found in two cases of osteosarcoma and one of Ewing's sarcoma, which is comparable to other limb-salvage series for malignant bone tumours [12]. Yet, with amputations, the long-term survival of these patients was unaltered indicating the need for vigilant surveillance in limb-salvage procedures.

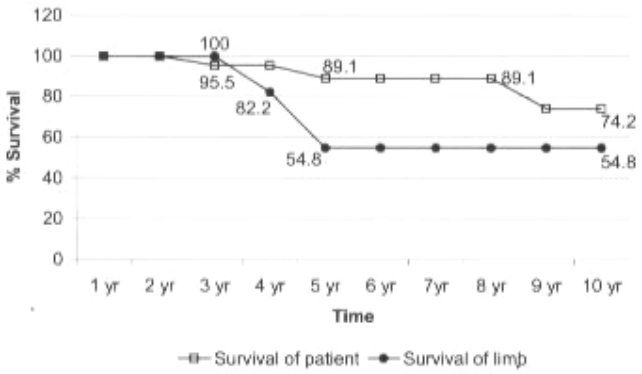

Fig. 1 Kaplan-Meier survivorship analysis showing 5- and 10-year survival of the patient and the limb in aggressive and malignant fibular tumours. *X* axis, time in years; *Y* axis, survival percentage

Biological complications in the form of superficial infections or flap necrosis were not major determinants of the functional outcome, as they were managed with appropriate measures. However, following wide resection in 16 patients of proximal third lesions, peroneal nerve palsy developed in two, one of whom required a permanent ankle-foot orthosis. Post-operative immobilisation resulted in fibrous adhesions that offered considerable stability in distal fibular resections without any need for ankle reconstruction [1, 9]. This was evident as both of our patients with distal fibulectomy had a near-normal ankle function, though one had roentgenographic evidence of lateral talar shift. The three radical resections, however, resulted in functional morbidity; the fact that two of them did not develop any adverse oncological outcome reiterates that such margins should be carried out for high-grade sarcomas. None of the three patients who died of disseminated disease developed local recurrence, a rate similar to that reported by Malawer [8]. This emphasises the need for aggressive chemotherapeutic control of the disease process even after providing adequate local oncological clearance.

Fibular resections when performed appropriately, irrespective of tumour site, and when adhering to strict oncological principles can provide disease-free survival rates comparable to those of amputations. The advantage of psychosocial and emotional acceptance and the ability to ambulate independently expands the scope of salvage surgeries for tumours of the fibula. Moreover, as the world-wide scenario of limb salvage has gathered considerable momentum, lower complication rates and higher functional scores have been reached. This series describes an effective salvage method of treating aggressive and malignant lesions of the fibula allowing for a useful limb while at the same time increasing the disease-free and long-term survival of the patient.

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