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Limb salvage surgery for pathological fractures in osteosarcoma

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Abstract The local treatment of pathological fracture in patients with a primary osteosarcoma remains controversial. In this paper we report the oncological outcome of the treatment of pathological fractures in 18 patients suffering from this disease. There were ten male and eight female patients, and the average age at diagnosis was 17 years. All patients received adjuvant chemotherapy. Wide resections were performed in 17, but in one there was 'contamination' of the margins of the excision. Skeletal reconstruction was performed with a locally designed and manufactured custom 'mega' prosthesis. The average follow-up was 33 months (range: 12–93 months), and 14 patients were alive on completion of the study. Local recurrence appeared in two patients, while three developed pulmonary metastases.

Résumé Le traitement local des patients ayant une fracture pathologique due à un ostéosarcome primaire reste sujet à controverse. Nous avons fait une étude sur le résultat oncologique de la chirurgie conservatrice chez 18 patients d'un âge moyen de 17 ans lors du diagnostic ayant des fractures pathologiques d'ostéosarcome. Tous les patients avaient subi d'une chimiothérapie adjuvante. Les marges de résection étaient larges chez 17 patients et contaminées chez un patient. La reconstruction squelettique a été accomplie avec des prothèses faites sur mesures conçues et fabriquées localement. Le suivi moyen a duré 33 mois (entre 12 et 93 mois). Quatorze patients étaient vivants à la fin de cette étude. La récidive locale s'est manifestée chez deux patients alors que trois patients ont été atteints de métastase pulmonaire.

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Introduction

Modern chemotherapy has revolutionised the management of patients with primary osteosarcoma of bone. Local treatment alone produces a survival rate of 5 years in 20% of patients [10, 12], but with the addition of effective chemotherapy this improves to 60% [9]. However, the local management of a pathological fracture through a primary bone sarcoma remains controversial and amputation has been the most common surgical treatment [7]. Simultaneous advances in chemotherapy, surgical and imaging techniques have encouraged preservation of the limb as the first choice of management [3, 8]. Preoperative chemotherapy produces shrinkage of the tumour and may also result in fracture union, but even so the effect of the fracture on the final outcome, and on the prognosis of patients with osteosarcoma, remains uncertain [10]. In this paper we report our experience in 18 patients of limb salvage surgery combined with chemotherapy as the management of a pathological fracture through a primary osteosarcoma.

Patients and methods

Between 1988 and 1999, 233 patients with a primary osteosarcoma underwent limb salvage surgery with replacement using a locally designed and custom-manufactured 'mega' prosthesis. There were ten male and eight female patients, and the average age was 17 years (range: 7–27 years). The most common site was in the distal femur (eight patients; e.g., Fig. 1), all the tumours were 'staged' and the patients were assessed with routine blood tests. CT scanning and more recently MRI were also used. Open biopsy was performed in all patients: 17 had a stage IIB tumour and one a stage IIIB tumour (Musculoskeletal Tumour Society Staging System [4]). The patient with a stage IIIB lesion also had a solitary pulmonary metastasis, which was resected. The criteria for selection of patients for limb salvage surgery was the same, whether there was an associated pathological fracture or not, and was based on the local extent of the tumour as determined by staging studies and by a good response to chemotherapy. Patients were excluded if they had an inappropriate biopsy scar, infection, tumour involvement of neuro-vascular structures, involvement of a joint, multiple pulmonary metastases or disseminated disease. All pa-

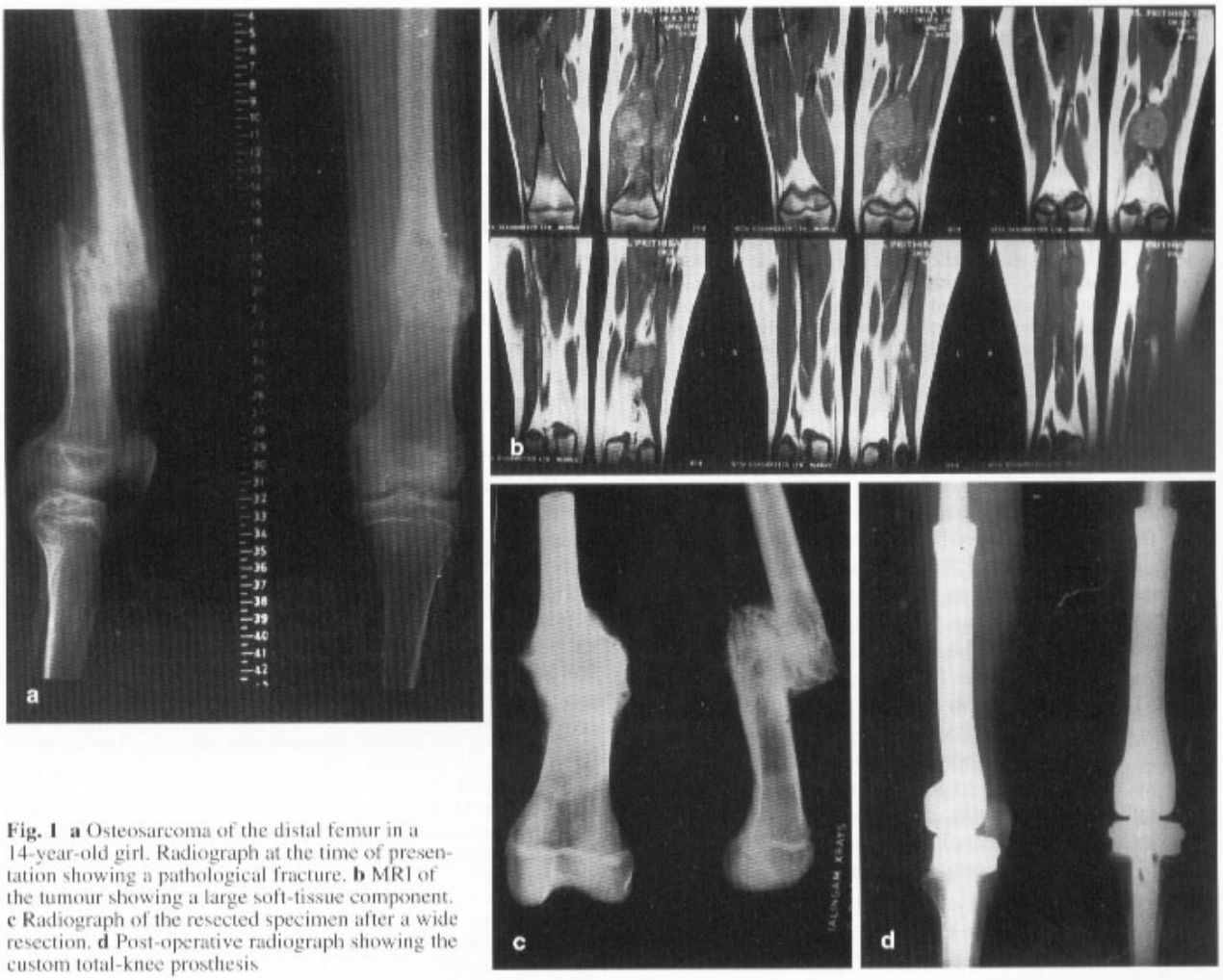

Fig. 1 **a** Osteosarcoma of the distal femur in a 14-year-old girl. Radiograph at the time of presentation showing a pathological fracture. **b** MRI of the tumour showing a large soft-tissue component. **c** Radiograph of the resected specimen after a wide resection. **d** Post-operative radiograph showing the custom total-knee prosthesis

Table 1 Details of 18 patients who underwent limb salvage surgery for pathological fractures in osteosarcoma (oncological results: *NED* no evidence of disease, *DOD* died of disease, *AWD* alive with disease)

Case no.	Anatomical site	Age (years)	Sex	Oncological result	Functional result	Local recurrence	Lung secondaries	Follow-up (months)
1	Distal femur	21	M	NED	Good	–	–	93
2	Distal femur	22	F	DOD	Fair	–	+	21
3	Distal femur	11	F	NED	Excellent	–	–	68
4	Proximal humerus	18	M	NED	Excellent	–	–	63
5	Distal femur	21	M	DOD	Good	–	+	24
6	Proximal tibia	17	F	NED	Good	–	–	49
7	Distal femur	15	M	NED	Good	–	–	48
8	Proximal femur	16	M	NED	Excellent	–	–	48
9	Distal femur	14	F	DOD	Good	–	+	1
10[a]	Proximal humerus	22	M	DOD	Poor	+	–	9
11	Proximal tibia	14	M	NED	Fair	–	–	34
12	Distal femur	14	M	NED	Good	–	–	32
13	Distal femur	27	M	NED	Fair	+	–	28
14	Shaft of femur	14	F	NED	Good	–	–	19
15	Shaft of femur	20	F	NED	Good	–	–	15
16	Proximal humerus	12	M	NED	Good	–	–	15
17[b]	Proximal humerus	21	F	AWD	Excellent	–	–	14
18	Proximal tibia	7	F	NED	Fair	–	–	12

[a] Contaminated margin of resection; in all other cases wide margins were achieved
[b] Stage IIIB disease; in all other cases stage IIB disease was present

tients received three courses of pre-operative chemotherapy, with an interval of 3 weeks between each course. Each of these consisted of Ifosfamide (6 mg/m^2 in separate doses over 3 days) combined with Mesna, Adriamycin (50 mg/m^2) and Cisplatin (100 mg/m^2 over 3 days). A wide margin of resection was achieved in 17 patients while in one there was evidence of contamination. The length of bone removed was 101–150 mm in six cases, 151–200 mm in seven, and >200 mm in five. Skeletal reconstruction after resection was done with a locally designed and custom-manufactured 'mega' prosthesis. After operation the patients received a further three courses of the same chemotherapy schedule. Follow-up examination was carried out at an average of 33 months (range: 12–93 months). The clinical data, the oncological and functional outcome and follow-up of our 18 patients who underwent limb salvage surgery for pathological fracture as a result of osteosarcoma is summarised in Table 1.

Results

In October 1999, when we last assessed our group of 18 patients, 14 (78%) had survived and of these 13 (72%) were free from disease while one patient was alive with disease. Four patients (22%) had died, three from pulmonary metastases and one as a result of local recurrence with disseminated disease. The functional result (using the 30-point rating system of the Musculoskeletal Tumour Society) was excellent in four (22%), good in nine (50%), fair in four (22%), and poor in one (6%). Two (11%) of the 18 patients developed local recurrence: one had been contaminated by 'spillage' of tumour material during a wide margin resection resulting in local recurrence, dissemination and death within 9 months, and the other patient with local recurrence was then treated by amputation. Three other patients (17%) died from pulmonary metastases.

Discussion

Pathological fracture through an osteosarcoma occurring before presentation, or during pre-operative treatment, is seen in 5–10% of patients [6]. They occur in high-grade primary malignant bone tumours either spontaneously or as a result of minimal trauma, due to their high cellularity, poor differentiation and loss of matrix. In our series, 7.7% had a pathological fracture at presentation, and the telangiectatic variety of osteosarcoma, which is highly vascular and largely osteolytic, is particularly prone to this complication [5].

A fracture through a primary bone tumour results in a haematoma which may spread and contaminate soft tissues, an adjacent neurovascular bundle or a joint, and in the past this usually resulted in amputation [6]. Abudu et al. [1] noted that amputation produced better eradication of the local tumour than limb salvage, but did not influence the overall survival. This has also been observed by other surgeons [10, 11]. The 5-year survival of all patients with a localised osteosarcoma is about 60% [8], while in our series 78% were alive after an average of 33 months. Limb-sparing surgery with adequate margins of resection can be achieved in many patients with a pathological fracture through a primary osteosarcoma without compromising their survival, but there is a significant risk of local recurrence. In their series Abudu et al. [1] found a local recurrence rate of 19% in patients with a pathological fracture treated by limb salvage while this did not happen in patients who underwent amputation. Ward et al. [12], analysing local recurrence following the surgical treatment of primary malignant bone tumours, reported a rate of 50% in pathological fractures as against only 13% when the 'bone' remained intact. In a study of 17 cases of bone sarcomas complicated by a pathological fracture, Delepine and Delepine [2] found two local recurrences in 13 survivors at 51 months follow-up (15%). They concluded that a pathological fracture should no longer call for amputation if adequate local resection is performed after appropriate conservative and chemotherapy treatment. In our series there was an 11% local recurrence rate in patients with a fracture, and this is the same percentage that we find if all our patients with osteosarcoma are included. Thus the risk of local recurrence in limb salvage surgery is not significantly higher in the presence of a pathological fracture. Satisfactory clear margins of resection can still be achieved in the majority of patients without compromising their survival.

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