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The management of pelvic chondrosarcoma in Japan

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Abstract A review of 135 patients with pelvic chondrosarcoma who had been treated at 58 institutions in Japan between 1989 and 1998. In this series ablative surgery was necessary in 14 patients, and a limb salvage procedure performed in 121 patients. The surgical margins were "intra-lesional" in 27 patients, "marginal" in 30, "wide" in 77 and "unspecified" in 1. Local recurrence occurred in 33 patients and distant metastases in 25. Post-operative infection occurred in 25 patients. Revision was performed in 10 patients. The oncological outcome was "disease free" in 92 patients, "alive with disease" in 17 and "dead because of the disease" in 26. The cumulative prospective 10-year survival rate for all patients was 65%. The 6 statistically significant prognostic factors that determined the outcome were: – surgical stage, site of tumour, size of tumour, surgical margin, functional mobility and activity level after surgery. Excision of the tumour with a wide margin, or with a wide margin with partly marginal areas, and subsequent stable reconstruction are essential for improving the results of pelvic chondrosarcoma treatment.

Résumé Une étude sur 135 patients atteints de chondrosarcome pelvien, qui ont été traités dans 58 établissements médicaux de tout le Japon au cours des dix dernières années, a été effectuée. Parmi eux, 14 patients ont subi une opération mutilante, et 121 un sauvetage du membre. Des marges chirurgicales ont été réalisées sous la forme "intralésion" pour 27 patients, "marginale" pour 30, "large" pour 77 et "équivoque" pour 1. Une récidive locale est apparue chez 33 patients, et des métastases chez 25. Une infection post-opératoire est apparue chez 25 patients. L'opération a été refaite sur 10 patients. Le résultat oncologique a été "non malade" pour 92 patients, "vivant mais malade" pour 17 et "mort de la maladie" pour 26. Le taux de survie cumulatif prospectif pour 10 ans a été de 65% pour l'ensemble des patients. 6 facteurs déterminant le résultat se sont révélés statistiquement: stade chirurgical, site de la tumeur, taille de la tumeur, marge chirurgicale, fonction des patients et niveau d'activité. L'excision de la tumeur avec une marge large ou avec une marge large avec zone marginale partielle, et la reconstruction stable s'avèrent essentielles pour améliorer le résultat du traitement du chondrosarcome pelvien.

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Introduction

Chondrosarcoma commonly occurs in the pelvis [15]. Because there are no major anatomical barriers to tumour extension in the pelvis, these sarcomas produce a large painful extra-skeletal mass but no other specific symptoms [13]. This has resulted in the treatment of pelvic chondrosarcoma (PCS) often involving surgical procedures that interfere with normal anatomy, and local and systemic recurrence and spread often results in a poor outcome. Previous authors have analysed the prognostic factors that determine the outcome of patients with PCS [13, 15, 17]. A review of PCS in terms of functional mobility, quality of life and survival rates was performed in order to observe the current trends in practice on a nation-wide basis, especially in terms of the prognostic factors.

Materials and methods

One hundred and thirty-five patients with pelvic chondrosarcoma treated at 58 institutions in Japan between 1989 and 1998 were assessed in this study. The patients were registered by members of the

Table 1 Data and results of this study

Value	Range (mean±SD)	Number (%)	*P*-value**	
Age at presentation	14–90 (48.2±15.8) Years			
Age at diagnosis	12–87 (44.3±15.9) Years			
Sex				
Male		73 (54%)		
Female		60 (44%)		
Unspecified		2 (2%)		
Method of biopsy				
Fine needle		1 (1%)		
Core		16 (12%)		
Open		95 (70%)		
Nil		22 (17%)		
Stage (Enneking)			0.0002	S
Stage IA		7 (5%)		
Stage IB		68 (50%)		
Stage IIA		7 (5%)		
Stage IIB		48 (36%)		
Stage IIIB		4 (3%)		
Unspecified		1 (1%)		
Site (reduplicative)*			0.0001	S
S-Sacrum		10		
I-Ilium (excluding acetabulum)		62		
II-Acetabular		54		
III-Ischio-pubis (excluding acetabulum)		44		
Size (maximal diameter)	3–43 (11.9±6.5) cm		0.0032	S
Associated conditions				
Olliers		2 (2%)		
Dyaphyseal aclasis (Osteochondromatosis)		27 (20%)		
Induction therapy				
Chemotherapy		12 (9%)		
Radiotherapy		3 (2%)		
SURGERY				
Margin intended (Enneking)				
Intralesional		9 (7%)		
Marginal		34 (25%)		
Wide		91 (67%)		
Unspecified		1 (1%)		
Margin achieved (Enneking)			0.0136	S
Intralesional		27 (20%)		
Marginal		30 (22%)		
Wide		77 (57%)		
Unspecified		1 (1%)		
Resection (reduplicative)*			0.1255	NS
SI ala+adjacent ilium		14		
I ilium		67		
Ia para acetabular+hip joint		48		
III ischio pubis		25		
IIIa ischio pubis+hip joint		17		
IIIp pubis only		11		
Reconstruction*			0.4438	NS
Nil		70		
Mega prosthesis		11		
Arthrodesis:				
ischio femoral		4		
ilio femoral		7		
autograft arthrodesis		5		
Massive allograft: without total hip		2		
Massive allograft: with total hip		2		
Autograft:				
vascularized		11		
not vascularized		24		
with total hip		10		
Composite: allo- & autograft		3		
Adjuvant therapy				
Irradiation		5		
extracorporeal		4		
Chemotherapy		8		

Table 1 (continued)

Value		Range (mean±SD)	Number (%)	*P*-value**	
Outcome					
Alive: disease free			92 (68%)		
with disease			17 (13%)		
Died (in month after surgery)			26 (19%)		
Local recurrence			33	0.0001	S
Distant metastases			25	0.0001	S
Function					
Patient (ISOLS)	Excellent		43 (32%)	0.0001	S
	Good		31 (23%)		
	Fair		28 (21%)		
	Poor		28 (21%)		
	Unknown		5 (3%)		
Prosthesis (ISOLS)	Excellent		2		
	Good		5		
	Fair		3		
	Poor		6		
Activity level				0.0001	S
Chairbound			20 (15%)		
Crutches			27 (20%)		
Stick			25 (18%)		
No walking aids			63 (47%)		
Infection			25 (19%)	0.3391	NS
Time in months post surgery		0–69 (9.0) months			
Revision			10 (7%)		
Time in months post 1st op.		2–59 (16.2) months			

SD: the standard deviation; NS: not significant; S: significant
* The number of patients is reduplicative
** P value is based on the oncological outcome, and expressed according to the Log-Rank test

Japanese Musculo-Skeletal Oncology Group (JMOG). The overall data collected is shown in Table 1. Operative treatment consisted of ablative surgery such as hemi-pelvectomy in 14 patients (10%) and a limb salvage procedure for 121 patients (90%). Reconstruction methods included prosthetic replacements in 23 patients, arthrodesis in 16 patients, autograft in 42 patinets, allograft in 5 patients, and 70 patients received no reconstructive surgery. Follow-up ranged from 3 months to 120 months (mean: 47.3 months). Patients were assessed in terms of oncological outcome, functional mobility, activity level, and complications. Functional mobility was expressed according to the International Symposium on Limb Salvage (ISOLS) scale, which is based on Enneking's criteria [3, 4]. In the statistical analysis the cumulative prospective of overall survival was calculated using the method of Kaplan-Meier [6]. The statistical significance of the differences between the survival curves was evaluated using the Log-Rank test and the generalised Wilcoxon test, with the criterion of probability being less than 0.05. The differences between the individual factors were assessed using the chi-squared test with the same criterion as for the above-mentioned tests. In addition the Cox's regression hazard model using the Stepwise method was used to rank the factors and to reveal prognostic values in the univariate analysis.

Results

The data available is shown in Table 1. The cumulative prospective 10 year survival rate for all patients was 65%. Those patients with a local recurrence or distant metastases had a worse oncological prognosis than those who did not (p=0.001).

In statistical analysis a significant difference (*P*=<0.05) based on the oncological outcome was shown for 6 factors:- surgical stage, site of the tumour, size of the tumour, the achieved surgical margin, functional mobility and activity level. Other factors including the method of reconstruction produced no significant difference in the oncological outcome (Table 1). When comparing the surgical stage with the oncological outcome, the survival rates in stages IA and IB showed a better prognosis than those in stages IIA and IIB (*P*=0.0002). However, the proportion of patients surviving in stage IB was less than that in stage IIIB; and the proportion in stage IIA who survived was also less than that in stages IIB and IIIB. In terms of the site of the tumour, patients with lesions smaller than 20 cm in diameter had better survival rates than those with tumours larger than 20 cm (*P*=0.0032). Failure to achieve a clear surgical margin resulted in a poor oncological outcome and the existence of an intra-lesional margin also resulted in a poor prognosis, although there was no significant difference between a wide margin and a slim margin (*P*=0.0136). In terms of functional mobility and level of activity, 75% of the patients in the "excellent" group survived, of those patients in the "good" group 93% survived, in the "fair" group 53%, and in the "poor" group 52% (*P*=0.0001). The survival rate in the "chairbound" group was 29%, in the "on crutches" group it was 71%, in the "walking stick" group it was 88%, and in the "no walking aids" group it was 72% (*P*=0.0001). With regard to reconstruction methods, the group receiving no reconstructive surgery had a good survival rate at 73%. The groups that underwent various reconstruction operations, however, had problems of instability or poor oncological out-

Table 2 Significance between the tumour site and functional mobility

	Excellent	Good	Fair	Poor	Total	χ^2
II – Acetabular	4	15	16	18	53	Significant
Others	39	16	12	10	77	$P<0.0001$
Total	43	31	28	28	130	

Table 3 Significance between the tumour site and activity level

	Chairbound	On crutches	Using a stick	No walking aid	Total	χ^2
II – Acetabular	12	16	12	14	54	Significant
Others	8	9	13	49	79	$P<0.0004$
Total	20	25	25	63	133	

IIa: Paracetabular+hip joint; IIIa: Ischio-pubis+hip joint

Table 4 Significance between the resectin area and functional mobility

	Excellent	Good	Fair	Poor	Total	χ^2
IIa and IIIa	4	17	20	20	51	Significant
Others	38	14	7	8	67	$P<0.0001$
Total	42	31	27	28	128	

Table 5 Significance between the resection area and activity level

	Chairbound	On crutches	Using a stick	No walking aid	Total	χ^2
IIa and IIIa	12	21	13	16	62	Significant
Others	8	92	12	46	68	$P<0.0001$
Total	20	23	25	62	130	

IIa: Paracetabular+hip joint; IIIa: Ischio-pubis+hip joint

comes, especially those who received a prosthesis or an arthrodesis ($P=0.4438$).

Apart from the oncological outcome, the statistical significance of the difference between the individual factors was: between the tumour site and functional mobility ($P<0.0001$) as shown in Table 2; between the tumour site and activity level ($P<0.0004$) as shown in Table 3; between the resection area and functional mobility ($P<0.0001$) as shown in Table 4; and between the resection area and activity level ($P<0.0001$) as shown in Table 5. There were no significant differences resulting from other factors. Tumours involving or near to the acetabulum (ISOLS II) were associated with poor functional mobility and level of activity. Resection in the paracetabular region (ISOLS IIa and IIIa) resulted in poor functional mobility.

The statistical significance of the difference (based on Cox's hazard model) is given for 7 criteria:- sex, surgical stage, "intra-lesional" partial excision, ilio-femoral arthrodesis, total hip replacement with autografting, a "chairbound" activity level, or an "on crutches" activity level ($P=0.0001$ using the chi-squared tests of –2 Log Likelihood, Score and Wald). Among the individual values recorded by the Wald test, 5 of the values (excluding sex and ilio-femoral arthrodesis) also showed statistically significant differences ($P<0.05$). Using the risk ratio the following results were statistically assessed:- the fatal risk of females was 0.457, that of surgical stages 2.135 in each stage, that of the intra-lesional margin 3.978; that of ischio-femoral arthrodesis 5.178, that of autografting with total hip replacement 5.576, that of "chairbound" 5.559, and that of "on crutches" 4.951.

Discussion

Chondrosarcoma is a relatively rare tumour [15], and this makes it difficult to find relevant data. However, our retrospective study may provide some insight into the behaviour and outcome of PCS, and although it was based on results from patients who had been treated at various hospitals, the difference in each surgeon's treatment strategy seemed to be minimised as a result of adequate discussion at various meetings held in Japan on musculo-skeletal oncology [7, 8, 9, 19].

Some authors [13, 15, 17] consider that the histological grade of the chondrosarcoma is of extreme importance as a prognostic factor, and this together with the

surgical stage was assessed in our research. Low-grade sarcomas in stages IA and IB showed better survival rates than high-grade sarcomas in stages IIA and IIB. However, the proportion of patients surviving in stage IB was less than that in stage IIIB, and that in stage IIA was also less likely to survive than those in stages IIB and IIIB. On the other hand, Pritchard et al. [15] suggest that tumour size could be of prognostic importance, patients with lesions smaller than 10 cm in diameter having a better prognosis than those with lesions larger than 10 cm. In contrast Ozaki et al. [13] stress that tumour size has no impact on the overall survival rate. In our series patients with tumours larger than 20 cm in diameter had a poor prognosis, and in addition the surgical stage, site of the tumour, resection area, achieved surgical margin, functional mobility and level of activity were found to be statistically important prognostic factors.

The size and site of the tumour, however, as well as the surgical stage are beyond the surgeon's control. The area of resection must be decided according to the site of the tumour while the functional mobility and activity level of the patient will depend on the technique and adequacy of reconstruction. For many surgeons, therefore, the most important factors for improving the outcome of patients with PCS are the 'achieved surgical margin' and the method of reconstruction. Complete excision of the tumour when possibly and subsequent appropriate reconstruction are both necessary to produce an excellent result and satisfactory function.

With regard to the 'achieved surgical margin' of the excised tumour in our series there was no significant difference between those removed with a wide margin and those removed with a slim margin, although an *intra-lesional margin* resulted in a poor prognosis. However, other authors note that a 'marginal margin' in their series was actually recorded as a wide margin with partly marginal area. Current margin assessment has been very strict in Japan since the introduction of Kawaguchi's method [7, 8] and therefore the authors would like to stress that achieving at excision either a wide margin or a wide margin with partly marginal area is essential in order to achieve local cure. This also ensures a safety margin in the removal of a pelvic chondrosarcoma.

In our series, the method of reconstruction did not statistically alter the oncological outcome. The patients who required reconstruction either had a huge sarcoma, or a tumour in the paracetabular region. PCS located in other parts of the ilium, pubis or ischium were simply excised. Various methods of reconstruction were used in a total of 65 patients (Table 1). In Japan massive allografting is not as often performed as in America or Europe because of religious beliefs and constraints. However, patients who underwent any reconstruction other than allo- or autografting had unexpected instability, poor oncological outcomes, and unsatisfactory functional mobility. It is possible that surgeons hesitated to resect a tumour radically in order to reduce the extent of the reconstruction required.

Many authors have devised and described their own original procedures for PCS patients suffering from complications [1, 2, 10–12, 14, 16–20]. A PCS located in the upper part of the ilium requires simple excision [2, 5]. When the sarcoma is in the paracetabular region there are options of resection arthroplasty, arthrodesis, or prosthetic replacement including "polyacetal hemipelvis" combined with allograft and/or autograft [1, 2, 9–11, 18, 19]. Based on previous articles and the research for our study, resection arthroplasty and arthrodesis produce stability with few complications. In contrast, prosthetic replacement is more likely to result in infection. Post-operative infection can be related to a large dead space and a haematoma [12]. The prevention of infection and a stable reconstruction are required to treat massive tissue defects after a radical and wide resection.

We have found that excellent oncological and functional results were only achieved for those tumours that were not located in the paracetabular region, as in these excision with wide margins coud be achieved easily, and massive reconstruction following tumour resection was unnecessary (Tables 2, 3, 4, 5). Moreover, the histological grading of the tumour was also an important prognostic factor. The problems of reconstruction after excision of PCS located in the paracetabular region have yet to be overcome.

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