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Abstract

We evaluated the surgical problems encountered during treatment of 14 patients with malignant tumors originating in the pelvic region at our department. The tumor involved the iliac bone in 6 patients, the ischial bone in 2, the pubic bone in 2, and the gluteal region in 4. Invasion to the sacrum was observed in 7 patients. Twelve patients underwent surgical procedures consisting of intralesional resection in 6, marginal resection in 3, and wide margin resection in 3. Six of the 7 patients with sacral invasion developed local recurrence. Two patients with chondrosarcoma and one with parosteal osteosarcoma survived for 4 or more years, but the mean survival period in those with high grade malignant tumors was 11 months. These findings indicate the difficulties encountered in the treatment of malignant pelvic tumors.

KEYWORDS: bone neoplasms, soft tissue neoplasms, malignant neoplasms, pelvis, surgical treatment

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Clinical Study of Malignant Tumors Originating in the Pelvic Region

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We evaluated the surgical problems encountered during treatment of 14 patients with malignant tumors originating in the pelvic region at our department. The tumor involved the iliac bone in 6 patients, the ischial bone in 2, the pubic bone in 2, and the gluteal region in 4. Invasion to the sacrum was observed in 7 patients. Twelve patients underwent surgical procedures consisting of intralesional resection in 6, marginal resection in 3, and wide margin resection in 3. Six of the 7 patients with sacral invasion developed local recurrence. Two patients with chondrosarcoma and one with parosteal osteosarcoma survived for 4 or more years, but the mean survival period in those with high grade malignant tumors was 11 months. These findings indicate the difficulties encountered in the treatment of malignant pelvic tumors.

Key words : bone neoplasms, soft tissue neoplasms, malignant neoplasms, pelvis, surgical treatment

The diagnosis of malignant tumors originating in the pelvic region is often delayed due to their anatomical location, and thus curative surgery at the time of definite diagnosis is difficult in many cases (1, 2). Treatment results are therefore often not satisfactory.

Recent advances in imaging techniques are thought to allow earlier diagnosis and preoperative determination of the appropriate operative resection margins (3, 4). In addition, adjuvant therapies such as intensive chemotherapy have been introduced (5, 6). However, new imaging techniques and adjuvant therapy can be investigated in only a few such patients, because such tumors are rare (we encounter at most 1 or 2 each year). In spite of the development of such

methods, the most important aspect of treatment of pelvic tumors is surgical en block resection, which, in the pelvic region, is difficult to perform for anatomical reasons. In the present study we evaluated therapeutic difficulties encountered in patients treated at our department between 1967 and 1990.

Subjects and Methods

The subjects consisted of 14 patients (7 males and 7 females) with malignant tumors originating in the pelvis (10 bone tumors and 4 soft tissue tumors) ranging in age from 11 to 47 years (mean age, 24.8 years). Patient characteristics are summarized in Table 1. Bone tumors were histologically diagnosed as 4 Ewing's sarcomas, 3 chondrosarcomas, and one each parosteal osteosarcoma,

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osteosarcoma, and malignant fibrous histiocytoma (MFH). The 4 soft tissue tumors consisted of 2 MFH, one synovial sarcoma, and one malignant schwannoma. The tumors were located in the iliac bone in 6 patients, ischial bone in 2, pubic bone in 2, and the gluteal region in 4. The mean follow-up period was 50.2 months. Invasion to the sacrum was observed in 3 patients with bone tumors and in all 4 patients with soft tissue tumors. According to Enneking's surgical staging system (7, 8), Stage IB was observed in 3 patients, Stage IIB in 9, and Stage III in 2 (Table 1).

Results

The initial symptom in many patients was pain in the lumbogluteal region or in the leg, a symptom which is similar to that in lumbar spinal disorders. Prior to use of CT scanning (in 1985), the mean interval between the appearance of symptoms and definite diagnosis was 10.8 months, but this interval was reduced to 4.1 months thereafter (Table 1).

Twelve patients underwent surgery (Table 2). Hemipelvectomy was performed in 5 patients and tumor resection in 7. Resection was intralesional in 6 patients, marginal in 3, and with a wide

margin in 3 (with cuts into the tumor in 4 cases; patients 9, 10, 13, and 14). Of the 5 hemipelvectomies, marginal resection was done in 2, and intralesional resection in 3. Surgery was not performed in two patients due to Ewing's sarcoma with lung metastasis at the time of definite diagnosis (patients 7 and 12) (Table 1). These patients were treated with chemotherapy alone. Intraoperative and postoperative complications consisted of massive bleeding (more than 5,000 ml) in 2 patients (patients 2 and 10), and posttransfusion hepatitis (patient 8), hemorrhagic cystitis (patient 10), and wound infection (patient 2).

Adjuvant therapy consisted of chemotherapy in the 3 patients with Ewing's sarcoma and in one with osteosarcoma, while irradiation was performed on two patients with Ewing's sarcoma, one with MFH, and one with parosteal osteosarcoma (Table 1). The resection margins were reduced in 2 patients with Ewing's sarcoma after these treatments (patients 11 and 13). Irradiation therapy markedly decreased the tumor size and probably prolonged survival period in one patient with recurrent MFH (patient 10).

Local recurrence was observed in 5 of 6 patients treated by intralesional resection alone, 2

Table 1 Characteristics of patients with malignant tumors originating in the pelvis

Patient no.	Diagnosis	Age (yrs.)	Sex	Site	Chief complaint	Surgical stage ^a	First visiting	Interval ^b (mos.)	CT	MRI	Chemotherapy	Irradiation therapy
1	Chondrosarcoma	16	M	Pubic bone	Leg pain	IIB ^c	1967	18				
2	MFH	16	M	Ischial bone	Buttock pain	IIB	1970	13				
3	Synovial sarcoma*	17	M	Buttock	Buttock pain	IIB	1973	5				
4	Parosteal osteosarcoma	44	F	Iliac bone	Buttock pain	IB	1976	12				60 Gy
5	Chondrosarcoma	42	F	Ischial bone	Coxalgia	IB ^d	1981	1				
6	Osteosarcoma	15	F	Iliac bone	Buttock pain	IIB	1982	3			HDMTX ^e	
7	Ewing's sarcoma	11	M	Iliac bone	Limping	III	1985	1	○		VACA ^f	60 Gy
8	Malignant schwannoma*	34	F	Buttock	Buttock pain	IIB	1985	6	○			
9	Chondrosarcoma	21	F	Pubic bone	Leg pain	IB ^d	1986	2	○			
10	MFH*	47	F	Buttock	Buttock pain	IIB	1988	6	○			
11	Ewing's sarcoma	19	M	Iliac bone	Buttock pain	IIB	1989	4	○	○	VACA ^f	50 Gy
12	Ewing's sarcoma	15	M	Iliac bone	Leg pain	III	1989	3	○	○		
13	Ewing's sarcoma	21	M	Iliac bone	Buttock pain	IIB	1990	7	○	○	VACA ^f	
14	MFH*	29	F	Buttock	Lumbago	IIB	1990	4	○	○		

a: Enneking's surgical stage (7, 8). b: The interval between the appearance of symptoms and definite diagnosis, c: low-grade chondrosarcoma (9), d: high-grade chondrosarcoma (9), e: high dose methotrexate (5), f: VACA regimen consisting of vincristine, actinomycin-D, cyclophosphamide, and adriamycin (6), *: soft tissue tumor.

Table 2 Treatment and outcome in 12 surgically-treated patients

Patient no.	Diagnosis	Site	Type of location ^a	Initial op ^b	Resection line	Sacral invasion	Local recurrence	Distant metastasis	Treatment outcome (Observation period)
1	Chondrosarcoma	P	III	Resection	IL	-	+	-	DOD (25 mos.)
2	MFH	Is	III	Resection	M	-	+	+	DOD (48 mos.)
3	Synovial sarcoma*	Bu	I-s	Hemipel	IL	+	+	-	DOD (16 mos.)
4	Parosteal osteosarcoma	Il	I-s	Resection	IL	+	+	-	DOD (108 mos.)
5	Chondrosarcoma	Is	III	Resection	IL	-	-	-	CDF (98 mos.)
6	Osteosarcoma	Il	I-s	Hemipel	M	+	+	+	DOD (7 mos.)
8	Malignant schwannoma*	Bu	I-s	Hemipel	IL	+	+	-	DOD (6 mos.)
9	Chondrosarcoma	P	III	Resection	M	-	-	-	CDF (48 mos.)
10	MHF*	Bu	I-s	Hemipel	IL	+	+	-	DOD (20 mos.)
11	Ewing's sarcoma	Il	I-s	Resection	W	+	-	+	DOD (18 mos.)
13	Ewing's sarcoma	Il	I	Resection	W	-	-	+	NED (19 mos.)
14	MFH*	Bu	I-s	Resection	W	+	+	-	DOD (9 mos.)

Op: Operation, Il: Iliac bone, P: Pubic bone, Is: Ischial bone, Bu.: buttock, hemipel: hemipelvectomy, M: marginal margin, W: wide margin, IL: intralesional margin, CDF: continuous disease free, NED: no evidence of disease, DOD: died of disease.
 a: type of tumor location (O'Connor's system) (10), b: surgical margin of operation (11), *: soft tissue tumor.

of 3 treated by marginal resection, and one of 3 treated with wide margin resection. Additional resection was performed twice in the patient with recurrent parosteal osteosarcoma (marginal, patient 4), once in one patient with chondrosarcoma (intralesional, patient 1), and once in one patient with MFH (marginal, patient 2). Distant metastasis occurred in 4 patients. Two patients with chondrosarcoma have survived the entire observation period without evidence of disease, and 3 survived for 4 years or more. At the end of the observation period, 3 patients were alive without disease and the mean survival period was 55 months (ranging from 19 to 108 months). Nine patients treated surgically died, with mean survival period of 31 months (Table 2): cause of death was local recurrence of tumor in 6 patients and respiratory failure due to lung metastasis in 3.

The 3 most interesting cases are presented below.

Patient 9. A 21-year-old woman with grade 1 chondrosarcoma of the right pubic bone (9). The patient consulted our hospital with a 4-month history of pain in the right thigh. Plain radiography showed an abnormal shadow in the right pubic bone (Fig. 1a). There has been no local recurrence in 4 years even though resection was marginal (Fig. 1b). Surgery, even if only intralesional resection, may be very important in

this disease, since no local recurrence was observed in 2 of the three patients with chondrosarcoma, all of whom were treated surgically.

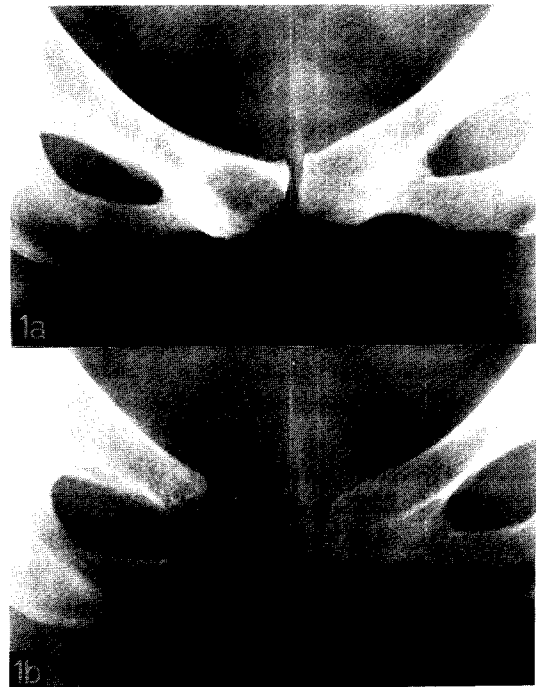


Fig. 1 Patient 1, Chondrosarcoma. (a) Plain radiograph at the time of initial consultation. (b) post operative plain radiograph.

Patient 13. A 21-year-old man with Ewing's sarcoma of the left iliac bone whose chief complaint was mild intermittent pain in the left gluteal region. Plain radiographs and MRI revealed a very large tumor in the iliac bone and gluteal region (Fig. 2a and 2b). Preoperative intensive chemotherapy consisting of adriamycin, vincristine, actinomycin-D, and cyclophosphamide was effective (Fig. 2c). After the left iliac bone as well as the iliac and gluteal muscle groups was extensively resected, the iliac bone was autoclaved and replaced in its original site, and the pelvic ring was reconstructed. Though lung metastasis appeared 6 months after the operation,

the metastatic lesion disappeared after intensive chemotherapy. No local recurrence has been identified in the 16 months since the operation (Fig. 2d).

Patient 14. A 29-year-old woman with MFH of the left gluteal region. The patient reported pain in the left gluteal region. Radiological examination revealed a lesion in the iliac bone (Fig. 3a). The tumor was resected with a wide margin to include the iliopsoas muscle, sacroiliac joint, and transverse process of the 5th lumbar spine with cut into the tumor (Fig. 3b), MRI clearly revealed the tumor invasion area to be from the iliac bone to the sacroiliac joint. This imaging

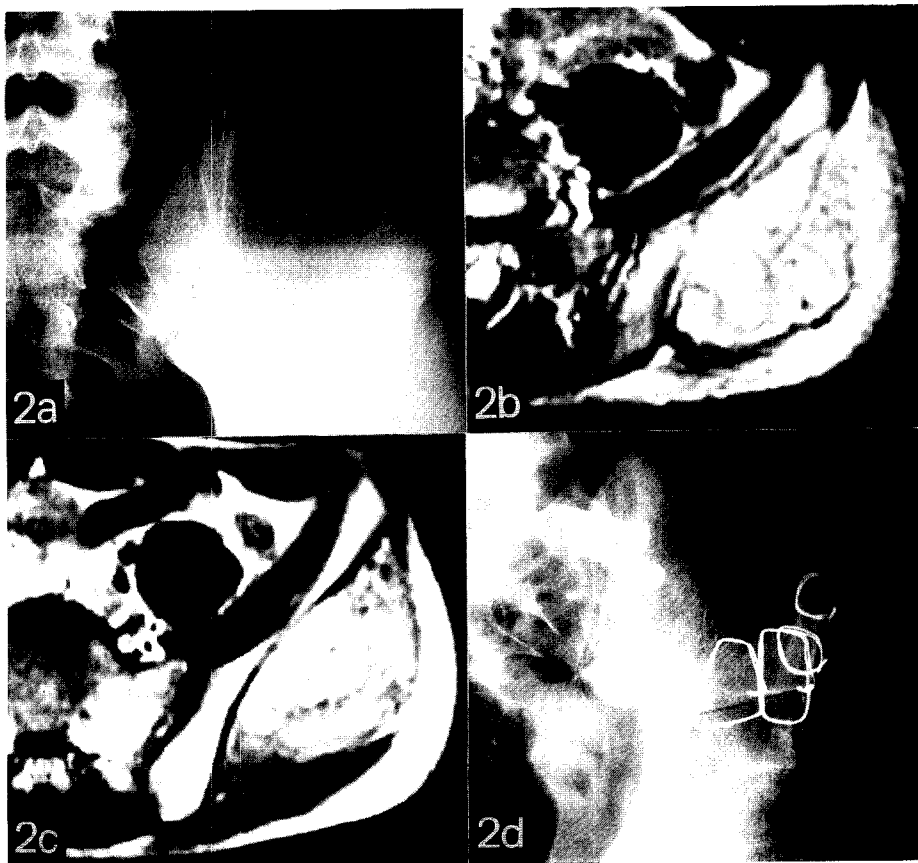


Fig. 2 Patient 14, Ewing's sarcoma. (a) Plain radiograph at the time of initial consultation. (b) MRI before preoperative chemotherapy (T2-weighted image). (c) MRI after preoperative chemotherapy (Gd-DTPA enhanced T1-weighted image). (d) Postoperative plain radiograph.

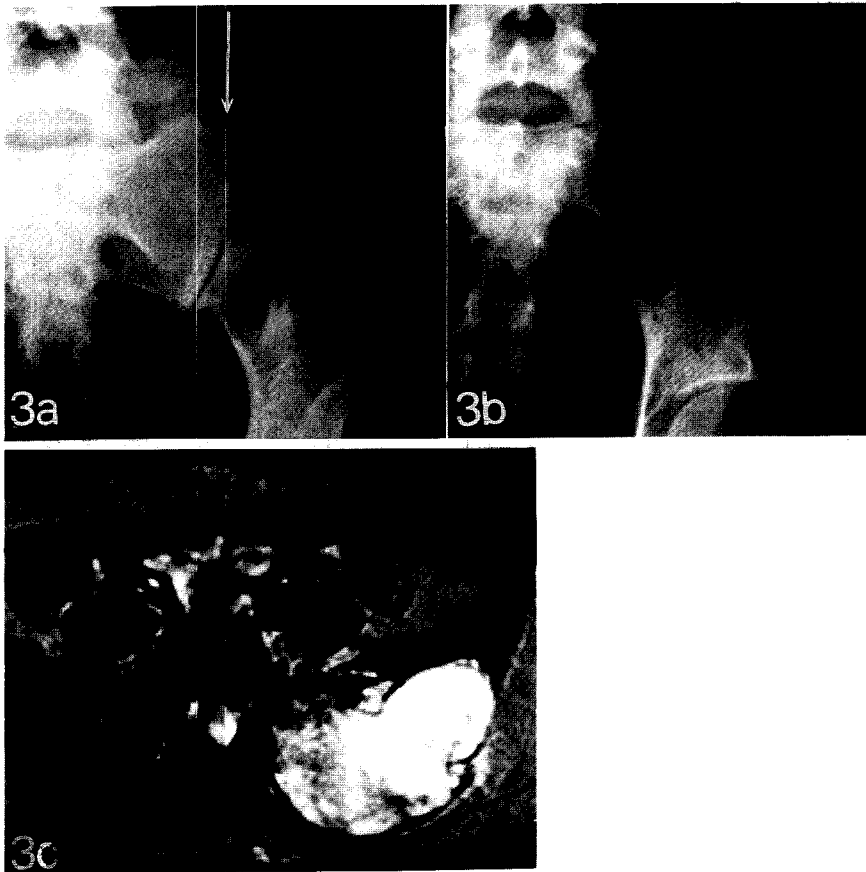


Fig. 3 Patient 13, MFH. (a) Preoperative plain radiograph. An arrow indicates bony destruction by the tumor. (b) Postoperative plain radiograph. (c) Preoperative MRI (T2-weighted image).

technique was useful for preoperative evaluation of the lesion (Fig. 3c). Local recurrence appeared in 4 months, and the patients died 9 months after the operation.

Discussion

Malignant pelvic tumors have been diagnosed earlier since the improvements in imaging techniques, but pelvic tumor is rarely suspected when lumbogluteal pain is encountered. Even if the pelvic tumor is detected and definite diagnosis is made, surgical treatment is difficult because of its

anatomical location. In all 9 of the surgically-treated patients who underwent intralesional or marginal resection, the tumors were very large, and in most cases had invaded the sacral bone. Six of the 7 patients with sacral invasion have had local recurrence. This finding suggests that local recurrence is greatly affected by the site of the tumor. All patients with sacral invasion were treated by hemipelvectomy and partial sacral resection, which effectively constituted only marginal or intralesional resection, and in 6 local recurrence was observed. Since there are many tissues at risk, the surgical procedures of necessity is often intralesional resection or marginal

resection, irrespective of whether the affected leg is amputated or preserved.

Invasion to the sacro-iliac joint is not taken into consideration in Enneking's classification of pelvic tumors (1, 2). However, O'Connor *et al.* have reported differences in the recurrence rate according to the tumor site, *e. g.*, 8% for type II (acetabular region), 17% for type III (ischiopubic region), 27% for type I (iliosacral region), and 38% with sacral or spinal invasion (type I-s) (10). Recurrence rate in our cases was 50% for type III and 100% for type I-s. Campanacci *et al.* evaluated local recurrence according to both tumor grade and resection margin, and found 12% recurrence at stage I (low grade malignancy), 43% at stage II (high grade malignancy), and 9% after wide margin resection, 27% after marginal or slight intralesional resection, and 66% after intralesional resection (12). In our cases, recurrence rate was 33% in low-grade malignancy, 72% in high-grade malignancy, 33% with wide margin resection, and 60% with marginal resection, and 83% with intralesional resection. Although the 2 patients with ischiopubic disease at Enneking's stage I were not treated with curative surgery, they have shown no local recurrence (patients 2 and 3). Thus, the local recurrence in our chondrosarcoma cases may also have been affected by tumor grade (7-9).

Reconstruction of the pelvic ring is sometimes necessary after resection of the pelvic region, especially in tumors around the sacro-iliac joint (13). Wide margin resection was performed in patient 13 (with Ewing's sarcoma), in whom preoperative chemotherapy using VACA was effective (6), and in whom reconstruction using the autoclaved bone as a spacer was possible (14). Incorporation and bone union between the grafted bone and host bone were often delayed because of the loss of activity of the bone morphologic protein, but this method was very useful because it is readily performed without reshaping the grafted bone.

Recent advances in diagnostic imaging tech-

niques, such as MRI, which is now extensively used, have allowed the routine determination of appropriate resection margins (Figs. 2b, 2c, and 3c) (3, 4). In recent years, bone and soft tissue tumors have often been treated by operation with preservation of the affected limb. For this operation, the resection margins should be preoperatively determined after accurate evaluation of the anatomical location of the tumor and invasion to the surrounding tissue. MRI affords high contrast resolution and provides images in various planes (15). All of 3 patients who underwent both operation and MRI could undergo resection with wide margin.

Considering the risk of recurrence at the resection margin in patients treated conventionally with hemipelvectomy, the number of cases in which other procedures, such as wide resection with preservation of affected limbs, can be used has theoretically increased (1, 2). Correct diagnosis of pelvic tumor was hastened and accurate determination of the line of resection improved as modern imaging technique and treatment for malignant tumors of the pelvic region gradually became established. However, wide resection or resection with a curative wide margin is very difficult because of the anatomical location, and is, therefore, performed in only a few patients. In the future, a curative operation may be possible if earlier diagnostic techniques, such as sensitive tumor markers, and/or more effective adjunctive method which allow reduction of the margin of resection, are developed.

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