



Surgical management of sacral chordoma

Harzem OZGER, Levent ERALP, Mustafa SUNGUR, Ata Can ATALAR

From Istanbul University Istanbul Faculty of Medicine, Istanbul, Turkey

Treatment results of 17 patients who were diagnosed with sacral chordoma between 1993 and 2007, were analyzed retrospectively. The mean duration of symptoms was 30.2 months. The mean tumour size was 10.7 cm ; the location was S2 or more proximal in ten-patients. A wide resection was achieved in 14 patients, a marginal resection in one patient and two patients had intralesional excision. Seven patients had a recurrence after a mean time interval of 36.3 months. Wound dehiscence and infection were the major problems. Four patients required continuous urinary catheterization because of incontinence, and ten patients had colostomy or ileostomy simultaneously with tumour resection. This study showed that tumour size and a wide surgical margin are importance factors for survival in chordoma patients. Use of a combined anterior and posterior approach could increase the chance of achieving a wide margin. A multidisciplinary approach is required to achieve this goal.

Keywords : sacrum ; tumours ; sacral chordoma.

INTRODUCTION

Chordoma is a rare, slow-growing, locally aggressive neoplasm of bone which arises from embryonic remnants of the notochord (4). Although rare, it is the most common tumour of the sacrum (4). Most cases are seen after the fifth decade of life. Of all chordoma cases, 50% are seen in the sacrum and coccyx, 30% in the skull base (sphenoccipital region) and 20% in vertebral bodies (4).

Chordomas are low- to intermediate-grade malignant lesions and are often diagnosed late because of indolent and vague symptoms and a location deep in the pelvis (10,20). Therefore, sacral chordomas are usually large at the time of diagnosis and they may involve adjacent neurovascular structures and vital organs in the pelvis. The tumour is usually Enneking stage IB at diagnosis, and its large size, the proximity to neurovascular structures and the congested pelvic structures make complete or wide tumour excision difficult. Larger and more proximally located tumours may require excision of sacral nerve roots leading to incontinence, sexual dysfunction and motor weakness (13).

Chordoma expands in the sacral foramina and plugs cephalad in the neural canal. Anteriorly, it is

-
- Harzem Ozger, MD, Professor of Orthopedics and Traumatology.
 - Levent Eralp, MD, Associate Professor of Orthopedics and Traumatology.
 - Mustafa Sungur, MD, Resident in Orthopedics and Traumatology).
 - Ata Can Atalar, MD, Specialist in Orthopedics and Traumatology.

Istanbul University, Faculty of Medicine, Department of Orthopedics and Traumatology.

Correspondence : Harzem Ozger, Istanbul University, Faculty of Medicine, Department of Orthopaedics and Traumatology, Millet Cad. Çapa, Fatih, Istanbul, Turkey.

E-mail : harzemo@yahoo.com

© 2010, Acta Orthopædica Belgica.

usually contained by the presacral fascia, so invasion of the rectal wall is not common. Posteriorly and laterally, it invades the gluteus maximus and piriformis muscles, sacroiliac joints, ligaments and subcutaneous fat (4). Metastases are not common and occur late in the course of the disease in the lung, bone, soft tissue, lymph node, liver and skin (8). The tumour has a delicate pseudocapsule, in which satellite lesions and occult extensions are frequent (4). Hence, local recurrence is common and usually occurs due to failure to achieve negative surgical margins. Therefore, achieving long term survival remains a challenge. Chordomas are insensitive to chemotherapy and also radioresistant (7). Therefore, complete surgical excision is the mainstay of treatment (6,10). Adequate surgical excision results in neurogenic dysfunction and pelvic instability, such that a radical surgical approach remains controversial (10).

The aim of the present study was to analyze the treatment results in a series of patients with sacrococcygeal chordoma to help to define the role of surgery and the relationship of tumour size, site, presentation type, approach and surgical margins to local recurrences and oncological outcome.

PATIENTS AND METHODS

All patients who were treated for sacrococcygeal chordoma at our institution between 1993 and 2007 were studied retrospectively. Seventeen patients (11 males, 6 females) who matched the required criteria were included in the study. Their average age at diagnosis was 55.5 years (range : 31-74) (table I).

Previous records of the patients were gathered retrospectively and presenting symptoms, duration of symptoms, size and site of the tumour, treatment method, surgical approach, complications, any recurrences and metastases were noted. The patients were examined for their last follow-up, and the duration of follow-up was calculated as the period from the first operation in our institution until last follow-up or death. The outcome was evaluated according to site, size, surgical approach and surgical margins.

Twelve patients initially presented at our institution without any prior operation and were diagnosed with sacrococcygeal chordoma after tru-cut biopsy following plain radiograph, CT and MRI of the involved region. Five patients presented secondarily to our institution.

Among those, one patient was diagnosed after incisional biopsy at another institution and was referred to us. One patient presented four months after receiving inadequate surgery with the intention of excisional biopsy. One patient had undergone tru-cut biopsy at another hospital but was diagnosed incorrectly with pleomorphic liposarcoma and had subsequently received chemotherapy and radiotherapy ; he was referred five months after the initial diagnosis. One patient presented with relapse 10 years after receiving suboptimal surgery and adjuvant radiotherapy. Finally, one patient presented with a third relapse after being operated inadequately in general surgery and neurosurgery departments at other institutions.

For surgical treatment, either a combined anterior and posterior approach or a single posterior approach was used. The surgical plan depended on the site and size of the tumour. If the tumour was larger than 10 cm in its greatest diameter, extended to the level of S2 and above, or invaded intrapelvic structures anterior to the presacral fascia irrespective of the site, a combined anteroposterior approach was used. If the tumour was smaller than 10 cm in its greatest diameter, and only involved the lower sacrum and posterior muscles, a posterior only approach was used. The aim of the surgery was to resect the lesion with safe wide margins. A wide margin was defined as a cuff of normal tissue around the tumour except anteriorly, where the tumour was covered by the presacral fascia. Therefore, it was separated easily from the rectum if there was no evidence of infiltration. If the presacral fascia or rectal adventitia were infiltrated, the rectum was included in the surgical resection. Proximally, according to the preoperative planning based on imaging techniques, at least 1-2 cm of normal bone was resected at the level of the osteotomy.

Eleven patients were operated with the combined anterior and posterior approach through the collaboration of the orthopaedic, general and plastic surgery departments (fig 1, a-c). Six patients were operated using a single posterior approach. The approach used also depended on our learning curve : among the six patients up to 2001, five were operated with a single posterior approach, whereas after 2001, this approach was used in only one of 11 patients, and the other 10 patients received the multidisciplinary and combined anterior and posterior approach. The combined anterior and posterior approach included anterior release and vascular exploration followed by rectum and/or sigmoid resection and colostomy by the general surgeon in eight patients and ileostomy in one patient ; vertical rectus abdominis myocutaneous flap (VRAM) preparation for dead space coverage by the plastic surgeon was done in five patients

Table I. — Patient Data

No.	Age	Gender	Symptoms	Duration of symptoms	Site	Size (cm)	Surgical approach	Margins	Additional Procedures	Radiotherapy	Saved nerve roots	Complications	Recurrence/metastases	Time to relapse	Follow-up	Outcome
1	44	m	pain, constipation, sciatalgia	16 months	S1-S4 sacrum + presacral region	12 × 8.5 × 6	P	Intra-lesional		yes	all saved	none	recurrence/none	39	118	DOD
2	53	m	pain, numbness	12 months	S2-S3	10.5 × 9	P	Marginal		yes	all saved	CSF fistula, urination/defecation problems, drop foot	recurrence/multiple bone metastases (81 mo)	77	81	DOD
3	59	f	pain, swelling	24 months	S3-caudal & anteriorly	15 × 11 × 10	P	Intra-lesional	FC-advancement flap from left gluteal region	yes	all saved		recurrence	36	48	DOD
4	48	m	pain, swelling	48 months	S3-caudal	8 × 7.5	P	WE		no	S1, S2, unilateral S3	transient anal incontinence, postop. cardio-pulmonary arrest	none/none	0	99	NED
5	63	m	pain, swelling	24 months	S2-coecyx	6 × 6 × 9	AP	WE		no	S1, S2 (Left)	pulmonary embolism, perioperative death	none/none	0	0	DOC
6	74	f	lumbar pain, swelling, and difficulty to sit	24 months	S4-caudal	16 × 14 × 12	P	WE		no	no sacrifice	surgical site infection, wound dehiscence	none/none	0	3	DOC
7	50	m	pain, swelling	16 months	S2-caudal	7 × 5	AP	WE		no	S1,S2	none	none/none	0	84	NED
8	57	m	pain, swelling	12 months	S3-coecyx	9 × 10 × 7 (rec)	AP	WE	sigmoid colon resection + colostomy (Miles)	yes	all roots resected	wound dehiscence	recurrence/-pulmonary metastasis (8 mo)	20	62	DOD
9	50	m	swelling	180 months	S2-coecyx	9 × 8 × 13.5	AP	WE	sigmoid and rectum excision, sigmoidostomy (Miles)	no	S1	difficulty in urination	recurrence /none	36	72	AWD
10	60	f	pain	24 months	S3, S4, left S5	6 × 11	AP	WE	ileostomy without rectum	no	L5	wound infection, meningitis, sepsis	none/none	0	3	DOD
11	65	m	pain	18 months	S3-caudal	10 × 2 × 4.2 (Third rec)	AP	WE	rectum resection + colostomy (Miles) + gluteus maximus flap	no	S1, S2 on right side and L5 on left side	drop foot, urinary incontinence, difficulty in ambulation	recurrence, pulmonary metastases (30 mo)	14	53	AWD
12	64	m	pain, constipation	4 months	S1-caudal	6 × 8.5	AP	WE	VRAM flap + rectum resection, colostomy (Miles)+ sacrum cryopreservation & reimplantation +lumbopelvic fixation	no	L4, L5	wound infection	recurrence /none	32	52	AWD

Table I. — Continuation

No.	Age	Gender	Symptoms	Duration of symptoms	Site	Size (cm)	Surgical approach	Margins	Additional Procedures	Radiotherapy	Saved nerve roots	Complications	Recurrence/metastases	Time to relapse	Follow-up	Outcome
13	45	f	pain	12 months	S1-S5	5 × 9 × 10	AP	WE	VRAM flap + sigmoid resection + colostomy without rectum (Miles)	no	–	perop. cardio-pulmonary arrest, exitus	none/none	0	0	DOD
14	52	m	pain	36 months	S4-coecyx	7 × 5	P	WE		no	all roots saved	none	none/none	0	37	NED
15	67	f	pain, constipation	36 months	S2,S3	8 × 7	AP	WE	VRAM + Miles, colostomy	no	S1	ARDS, Wound infection, wound necrosis	none/none	0	30	NED
16	31	f	pain, swelling, constipation	12 months	S2+right distal half sacrum	13 × 7.5 × 10.5	AP	WE	VRAM + sigmoid resection, colostomy without rectum (Miles)	no	S1	none	none/none	0	35	NED
17	61	m	pain	16 months	S2-coocyx	12 × 15	AP	WE	sigmoid excision, colostomy (Miles) + local flap	no	–	perop. exitus	none/none	0	0	DOD

FC : fasciocutaneous ; WE : wide excision ; VRAM : Vertical rectus abdominis myocutaneous flap ; DOD : dead of disease ; DOC : dead of complication ; AWD : alive with disease ; NED : no evidence of disease ; AP : combined anteroposterior approach ; P : posterior approach.



Fig. 1. — Sacrum chordoma in a 31-year-old female patient treated with the combined anterior and posterior approach. a : sagittal MRI section, tumour size is greater than 10 cm, and S2 is involved ; b : after the sacrum is prepared, a vertical rectus abdominis myocutaneous flap (VRAM) is harvested, a colostomy is performed with the anterior approach, and the tumour is resected with the sigmoid using the posterior approach ; c : clinical situation 10 months postoperatively.

(fig 2, a-d). To cover dead space, other techniques included a gluteus maximus advancement flap in two patients, a prolene mesh in five patients and a silicone breast prosthesis in two patients. Adjuvant radiotherapy was used in the first three patients in the series, who were treated with the posterior approach and had inadequate surgical margins. Two patients underwent total sacrectomy. One of those underwent total sacrectomy with the combined anterior and posterior approach followed by reconstruct-

tion. Rectum resection, ileostomy and harvesting of the VRAM flap was performed using the anterior approach and total sacrectomy, and laminectomy with preservation of the L4-5 roots was performed with the posterior approach. After resection was completed, the resected sacrum was treated with liquid nitrogen (cryopreservation) and reimplanted, and posterior instrumentation was performed after reinforcement with an antibiotic-loaded allomatrix and autograft application. Finally, the dead space was covered with the VRAM flap. The other patient who underwent total sacrectomy died perioperatively after resection of the sacrum, and thus the planned reconstruction process could not be performed.

The mean blood transfusion during the combined anterior and posterior approach was nine units of whole blood (range : 6-13 units) before 2004 ; the use of whole blood was subsequently discontinued due to changes in blood transfusion policy. After 2004, the mean blood transfusion was 19 units (range : 14-21 units) of erythrocyte suspension.

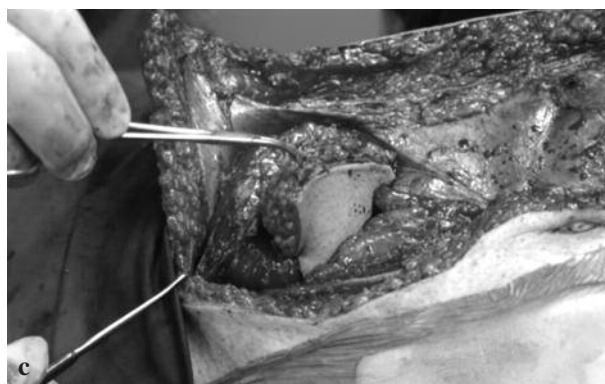


Fig. 2. — The preparation of a vertical rectus abdominis myocutaneous flap (VRAM) is shown. a : planning of the skin incision ; b : rectus abdominis is elevated ; c : flap is passed to the posterior incision ; d : postoperative image.

RESULTS

Pain was present in 16 patients and was the major presenting symptom. The second most common symptom was swelling (nine patients), followed by constipation (four patients) and sciatalgia and numbness (two patients). The main duration of symptoms before the patient was seen by any doctor was 30.2 months (4-180 months).

The tumour was localized below S2 in seven patients and at S2 or above in 10 patients. The mean greatest diameter of the tumour was 10.7 cm (range : 7-16 cm). The tumour was smaller than

10 cm in six patients and 10 cm or above in 11 patients.

Among patients for whom the posterior only approach was used, intralesional excision was achieved in two patients, marginal resection in one patient, and wide resection in three patients. Wide surgical margins were achieved in all 11 patients who were operated on with the combined anterior and posterior approach (tables II, III).

To achieve wide margins, some sacral nerve roots were sacrificed. However, all sacral nerve roots were saved in four patients who were treated with the posterior approach with intralesional, marginal

Table II. — Association of surgical approach with outcome

Approach	Number of patients	Surgical margins	Recurrence	Outcome
Combined anterior & posterior	11	wide	4 (36%)	6 alive (55%) ; follow-up : 54.3 mo. 5 dead (45%) ; 3 perioperative deaths at day 0, 1 DOC at 3 months, 1 DOD at 62 mo.
posterior	6	wide (3 pts), marginal (1 pt), intralesional (2 pts)	3 (50%)	2 alive (33.3%) ; follow-up : 68 mo. 4 dead (66.6%) ; 1 DOC at 3 mo., 3 DOD at 82,3 mo.

DOD : dead of disease ; DOC : dead of complication ; AWD : alive with disease ; NED : no evidence of disease.

Table III. — Association of surgical margins with recurrence

Margin	No. of patients	Approach	Recurrence	Time to recurrence
Wide	14	Anteroposterior (11), posterior (3)	4 (31%)	25.5 mo. (14-36)
Marginal	1	posterior	1 (100%)	77 mo.
Intralesional	2	posterior	2 (100%)	37.5 mo. (36-39)
Total	17		7 (41%)	36.3 mo.

or contaminated wide margins. In patients who underwent combined anterior and posterior treatment, S2 was saved in three patients, and S1 was saved in three patients. Due to nerve root sacrifice, urinary incontinence occurred in four patients, anal incontinence (excluding patients having a colostomy) in two patients, and drop foot in two patients. Early wound infection occurred in five patients.

Among the five patients who presented secondarily at our institution, three had a local relapse. Of the twelve patients who initially presented at our clinic, four had recurrence (table IV). Overall, seven patients (41%) had relapses after a mean duration of 36.3 months (14-77) (tables V-VI). Among those, three patients had received the posterior approach treatment with inadequate margins, and four were treated with the combined anterior and posterior approach. Among the latter, two patients had presented to our institution with relapses. One patient, who presented after incisional biopsy, and another patient, who was treated with cryopreservation and lumbopelvic fixation, also had relapses. For treatment of relapses, three patients underwent re-resection, and the other patients received only pain treatment because the lesions

were inoperable due to their site, size and multifocal nature. Three patients (18%) had metastasis after a mean duration of 39.7 months (8-81 months) : one with multiple bone metastases and two with pulmonary metastasis.

Two patients died intraoperatively due to cardiopulmonary arrest, and one patient died on the first postoperative day due to a pulmonary embolism. Excluding perioperative mortality, the mean follow-up was 55.5 months (3-118 months), with two patients dying three months postoperatively due to wound infection and sepsis (died of complication – DOC). Four patients died of disease (DOD) at a mean of 77.3 months (62-118). At their last follow-up, five patients had no evidence of disease (NED), and three patients were alive with disease (AWD). The overall survival rate in this study is 47% (eight alive and nine dead) at a mean follow-up of 55.5 months. The five-year survival rate is 43%.

DISCUSSION

Treatment of sacral chordoma requires aggressive surgery and is complicated by tumour biology, anatomical localization of the tumour, and late

Table IV. — Association of presentation type with outcome

Presentation	No. of patients	Recurrence	Outcome
Primary	12	4 (33%)	5 alive – 4 NED – 1 AWD 7 dead – 5 DOD – 2 DOC
Secondary	5	3 (60%)	3 alive – 1 NED – 2 AWD 2 dead – 2 DOD

DOD : dead of disease ; DOC : dead of complication ; AWD : alive with disease ; NED : no evidence of disease.

Table V. — Association of tumour size with outcome

Size	Approach	Surgical margin	Recurrence	Outcome
≥ 10 cm	4 posterior	2 intralesional, 1 marginal, 1 wide	5 (45.5%)	3 alive – 1 NED – 2 AWD 8 dead – 7 DOD – 1 DOC)
	7 anteroposterior	7 wide		
< 10 cm	2 posterior	2 wide	1 (16.7%)	5 alive – 4 NED – 1 AWD 1 dead – 1 DOC
	4 anteroposterior	4 wide		

DOD : dead of disease ; DOC : dead of complication ; AWD : alive with disease ; NED : no evidence of disease.

Table VI. — Association of tumour site with outcome

Site	Approach	Surgical margin	Recurrence	Outcome
S2 and above	2 posterior	1 intralesional, 1 marginal	4 (40%)	5 alive – 2 AWD – 3 NED 5 dead – 4 DOD – 1 DOC
	8 anteroposterior	8 wide		
Below S2	4 posterior	1 intralesional, 1 contaminated wide, 1 wide	3 (42.9%) (2 pts had presented with relapse)	3 alive – 2 NED – 1 AWD 4 dead – 3 DOD – 1 DOC
		3 anteroposterior		

DOD : dead of disease ; DOC : dead of complication ; AWD : alive with disease ; NED : no evidence of disease.

presentation of the patient. Chordoma is a low-grade malignant bone tumour, and it is thus resistant to radiotherapy and chemotherapy ; however, the tendency to local recurrence is high (5). Recurrence is inevitable in the case of suboptimal surgery, and frequent relapses eventually render the lesion inoperable. During such a course, bowel, urinary and sexual functions that are supposedly protected by performing suboptimal surgery are affected by the tumour itself. Functional loss and a huge tumour mass, sometimes to such a degree that the tumour protrudes from the skin, lead the patient to experience a long and painful terminal disease period. The only reasonable treatment method is tumour resection with adequate margins (10,11,12). If the surgeon and patient decide on an inappropriate operation to protect function and psychological health, the result could be worse than no treatment.

The sacrum has a complex anatomy due to its close relationship with major vessels, sacroiliac joints, sacral nerve roots and viscera like the bowel and rectum. This makes it hard to achieve wide surgical margins during surgery and increases perioperative morbidity and mortality. Resection of the tumour with wide margins may require sacrificing important nerve roots, mechanical elements that contribute to the stability of the pelvis and even viscera like the bowels and bladder, causing deficits in mechanical, neurological or visceral functions that may or may not be compensated for ; mechanical functions may be maintained with complex reconstructive methods, but bowel, bladder and sexual dysfunction is permanent due to the sacrifice of sacral nerve roots. It is essential that the patient be informed of all these risks and accepts them before treatment.

Chordoma may go unnoticed for a long time because the pelvis provides a large space for tumour growth. The mean time interval between the onset of symptoms and presentation was 30.2 months in this study ; it has previously been reported as 24 to 44.4 months in literature (3,10,13,17). In addition, when the symptoms become evident, the patient may present to the general surgery department for defecation problems, the urology department for urinary problems, or the neurology department for numbness, and these inadequate consultations will

delay treatment. Five patients in this study (33%) presented secondarily to our institution ; they were either treated initially or had undergone invasive diagnostic procedures at other centers. The recurrence rate was 60% for secondary presentation and 33% for patients who first presented at our institution. The patient may be reluctant to undergo treatment after learning about the risks of the operation and may even pressure the surgeon to perform insufficient surgery. The key in chordoma treatment is in briefing the patient about the nature of the disease, required treatment, expectations and prognosis.

The most important element in successful treatment is the surgeon. The surgeon may be uninformed about the pathology and natural course of a rare tumour such as chordoma. He/she may also lack experience, skills or the team for achieving wide margins in such a complex anatomy. He/she may also refrain from suggesting and applying the required surgical operation because of the morbidity and possible complications. Therefore, it is essential that the patient seek the best care, which includes a multidisciplinary team with an experienced oncologic orthopaedist who is familiar with the tumour biology and behaviour, a spine surgeon, a plastic surgeon and a vascular surgeon when necessary.

The mean tumour diameter in this study (10.7 cm) was slightly larger than reported in previous studies. Fuchs *et al* reported a mean diameter of 9 cm in a series of 52 patients (10) and Osaka *et al* reported 7.8 cm in a series of 12 patients (17). The rate of tumour recurrence was 45.5% if the maximum tumour diameter was 10 cm or above and 16.7% if it was less than 10 cm. Likewise, 27% of patients were alive at the last follow-up if the tumour diameter was 10 cm or above, and 83% were alive if it was less than 10 cm initially. A smaller tumour size is a favourable factor with respect to recurrence and survival, but a statistical analysis was not possible because of the limited number of patients.

The tumour extended to or above the level of S2 in 10 patients. Among those, two patients who were operated on early in the series were treated with the posterior approach with intralesional or marginal

resection, and both subsequently had recurrence. Eight patients were operated on with the combined anteroposterior approach and wide margins; only two of those patients recurred. Overall, 40% of patients with a tumour at higher sacral levels had recurrence. The tumour was located below the S2 level in seven patients. Four of those patients were treated with the posterior approach, and three patients were treated with the combined anterior and posterior approach; 42.9% of patients who had a tumour located below S2 experienced recurrence. The recurrence rate and survival do not appear to be influenced by tumour localization according to this study. However, in a previous study, the most proximal extent of the tumour was found to be the only significant independent factor in lumbosacral chordoma (7). The tumour extension was also important for perioperative risk and functional outcome. Resections proximal to S2 were significantly related with more blood loss, a longer ICU stay, higher hospital costs and more major complications (9). The risks of nerve sacrifice have also been studied. If the S3 nerve root is preserved unilaterally, bowel and bladder functions are most likely preserved. If S2 roots are sacrificed bilaterally, the bowel and bladder functions are usually lost, and the patient may need a colostomy because of compromised bowel functions (9). If the resection level is between the S1 and S2 levels, pelvic ring stability is compromised by 30%. If the resection passes through the S1 body, 50% of mechanical stability is lost, and lumbopelvic fixation is required (9,16). One patient in our series required lumbopelvic fixation after total sacrectomy, cryopreservation of the resected specimen and reimplantation of the cryopreserved bone with allograft.

The combined anteroposterior approach was used in 11 patients, and wide margins were obtained in all. The posterior approach was used in six patients; wide resection was achieved in three patients, marginal resection in one patient and intralesional excision in two patients. The recurrence rate was 36% for patients who had undergone the combined anterior and posterior approach and 50% for the posterior approach. At last control, 55% of the patients in the combined anterior and posterior approach group were alive, while only

33.3% of patients were alive if they had a single posterior approach. The patients who had undergone surgery with the posterior only approach were early patients in this series, treated before 2001, except one patient whose tumour was below the S4 level and smaller than 10 cm. The inadequate margins and high recurrence rate of the early patients urged our institution to reevaluate the treatment protocol, and it was concluded that a more radical surgery to obtain wide margins is essential in the treatment. This also conforms with the literature; complete resection and sacrifice of nerve roots if necessary at the time of surgery is advocated, and it is stressed that local failure rates are high for patients treated for recurrent chordoma (2,8,12). Bergh *et al* found that the recurrence rate was 17% for wide resections and 81% for intralesional or marginal resections (3). Ahmed found that ventrally, marginal resection is adequate due to the presacral fascia, which acts as a barrier, but posterolaterally wide margins are required to achieve lower recurrence and higher survival rates (1). The posterior approach is also advocated in the literature. Waisman *et al* recommended the posterior approach and mersilene mesh for sacral chordoma without local spread (19), but their series mostly included tumours at lower sacral levels.

The reported recurrence rate ranges from 70% to 33% in different studies (1,3,8,11). The recurrence rate in this study is 41%. Five-year survival as high as 86% has been reported (1,3,7,8,11,17). The mean follow-up in this series was 55.5 months, and the 5-year survival rate was estimated to be 43%. Nine patients were dead at last control: two patients died intraoperatively due to cardiopulmonary arrest; one patient died on the operation day after a massive pulmonary embolism; two patients died due to septic complications at three months; and four patients died of the disease at a median of 77.3 months (62-118 months). The early mortality at three months was hence 29.4%. The patients who died of disease (DOD) constituted 33% of cases in the series (four cases out of 12; DOD in a mean of 77.3 months), excluding patients who died perioperatively and in the early postoperative period due to complications. In one study, five out of 16 patients (31%) were reported as DOD after 31.4 months (12), which

agrees with our study ; however, considering the survival rates reported in the current literature, the mortality rate in this study is still high. Nevertheless, the studies in the literature include a variety of patients and surgical methods, and meta-analyses that assess studies conducted on similar patients and similar treatments are required to evaluate the survival rate of sacral chordoma.

Metastases occurred in three patients (3%) in this series after a mean time interval of 39.7 months (8-81 months). Two patients died of disease, and one patient was alive with disease at last control. The rate of metastasis is between 20% and 43% in literature (4,8).

The videolaparoscopic anterior approach is a novel technique with low morbidity, and has been utilized in recent years (14,15,18). In this series, we performed either a colostomy and/or VRAM flap in patients requiring an AP approach ; thus, an endoscopic anterior dissection was technically not applicable.

CONCLUSION

This study showed that a wide surgical margin and tumour size are importance factors for survival in chordoma patients. Despite the use of wide excisions and extensive application of the combined anterior and posterior approach, frequent recurrence is expected. The combined anterior and posterior approach could increase the chance of achieving a wide margin. If the surgeon does not sacrifice the nerve roots and their functions, the tumour will do so in time, but with an increased mortality. For sacral chordomas, which are low grade malignancies, the only chance for cure is a wide resection that includes whatever structure is invaded by the tumour. Complex spinal and plastic reconstructions are of vital help. A multidisciplinary approach lead by an oncologic orthopaedist is required to achieve this goal.

REFERENCES

1. **Ahmed AR.** Safety margins in resection of sacral chordoma : analysis of 18 patients. *Arch Orthop Trauma Surg* 2009 ; 129 : 483-487.
2. **Atalar H, Selek H, Yıldız Y.** Management of sacrococcygeal chordomas. *Int Orthop* 2006 ; 30 : 514-518.
3. **Bergh P, Kindblom LG, Gunterberg B, Kindblom LG.** Prognostic factors in chordoma of the sacrum and mobile spine, a study of 39 patients. *Cancer* 2000 ; 88 : 2122-2134.
4. **Campanacci M.** Chordoma. In : Campanacci M (ed). *Bone and Soft Tissue Tumors*. Springer Verlag, New York, Wien, 1999, pp 689-706.
5. **Casali PG, Stacchiotti S, Sangalli C Olmi P, Gronchi A.** Chordoma. *Curr Opin Oncol* 2007 ; 19 : 367-370.
6. **Chandawarkar RY.** Sacrococcygeal chordoma : review of 50 consecutive patients. *World J Surg* 1996 ; 20 : 717-719.
7. **Cheng E, Ozerdemoglu RA, Transfeldt EE, Thompson RC Jr.** Lumbosacral chordoma, prognostic factors and treatment. *Spine* 1999 ; 24 : 1639-1645.
8. **Chugh R, Tawbi H, Lucas D et al.** Chordoma : the non-sarcoma primary bone tumor. *Oncologist* 2007 ; 12 : 1344-1350.
9. **Devin C, Chong P, Holt GE et al.** Level-adjusted peri-operative risk of sacral amputations. *J Surg Oncol* 2006 ; 94 : 173-174.
10. **Fuchs B, Dickey ID, Yaszemski MJ, Inwards CY, Sim FH.** Operative management of sacral chordoma. *J Bone Joint Surg* 2005 ; 87-A : 2211-2216.
11. **Hanna SA, Briggs TWR, Cannon SR.** Sacral chordoma, can local recurrence after sacrectomy be predicted ? *Clin Orthop Rel Res* 2008 ; 466 : 2217-2223.
12. **Hulen CA, Temple HT, Fox WP et al.** Oncologic and functional outcome following sacrectomy for sacral chordoma. *J Bone Joint Surg* 2006 ; 88-A : 1532-1539.
13. **Jeys L, Gibbins R, Evans G, Grimer R.** Sacral chordoma : a diagnosis not to be sat on ? *Int Orthop* 2008 ; 32 : 269-272.
14. **Konstantinidis K, Theodoropoulos GE, Sambalis G et al.** Laparoscopic resection of presacral schwannomas. *Surg Laparosc Endosc Percutan Tech* 2005 ; 15 : 302-304.
15. **Lee KH, Tam YH, Chan KW et al.** Laparoscopic-assisted excision of sacrococcygeal teratoma in children. *J Laparoendosc Adv Surg Tech A* 2008 ; 18 : 296-301.
16. **Mindell ER.** Risks of sacral amputations. *J Surg Oncol* 2006 ; 94 : 173-174.
17. **Osaka S, Kodoh O, Sugita H et al.** Clinical significance of a wide excision policy for sacrococcygeal chordoma. *J Cancer Res Clin Oncol* 2006 ; 132 : 213-218.
18. **Tanaka J, Ito M, Shindo Y, Kotanagi H, Koyama K.** Laparoscopically assisted resection of the lower rectum. *Surg Endosc* 1996 ; 10 : 338-340.
19. **Waisman M, Kligman M, Roffman M.** Posterior approach for radical excision of sacral chordoma. *Int Orthop* 1997 ; 21 : 181-184.
20. **Wuisman P, Lieshout O, Sugihara S, van Dijk M.** Total sacrectomy and reconstruction, oncologic and functional outcome. *Clin Orthop Rel Res* 2000 ; 381 : 192-203.