Surgical Margins and Local Control in Resection of Sacral Chordomas

Pietro Ruggieri MD, PhD, Andrea Angelini MD,
Giuseppe Ussia MD, Maurizio Montalti MD,
Mario Mercuri MD

Published online: 16 July 2010
© The Association of Bone and Joint Surgeons® 2010

Abstract

Background The treatment of choice in sacral chordoma is surgical resection, although the risk of local recurrence and metastasis remains high. The quality of surgical margins obtained at initial surgery is the primary factor to improve survival reducing the risk of local recurrence, but proximal sacral resections are associated with substantial perioperative morbidity.

Questions/purposes We considered survivorship related to local recurrence in terms of surgical margins, level of resection, and previous surgery.

Methods We retrospectively reviewed 56 patients with sacral chordomas treated with surgical resection. Thirty-seven were resected above S3 by a combined anterior and posterior approach and 19 at or below S3 by a posterior approach. Nine of these had had previous intralesional surgery elsewhere. The minimum followup was 3 years (mean, 9.5 years; range, 3–28 years).

Results Overall survival was 97% at 5 years, 71% at 10 years, and 47% at 15 years. Survivorship to local recurrence was 65% at 5 years and 52% at 10 years. Thirty percent of patients developed metastases. Wide margins

were associated with increased survivorship to local recurrence. We found no differences in local recurrence between wide and wide-contaminated margins (that is, if the tumor or its pseudocapsule was exposed intraoperatively, but further tissue was removed to achieve wide margins). Previous intralesional surgery was associated with an increased local recurrence rate. We observed no differences in the recurrence rate in resections above S3 or at and below S3.

Conclusions Surgical margins affect the risk of local recurrence. Previous intralesional surgery was associated with a higher rate of local recurrence. Intraoperative contamination did not affect the risk of local recurrence when wide margins were subsequently attained.

Level of Evidence Level IV, therapeutic study. See Guidelines for Authors for a complete description of levels of evidence.

Introduction

Chordomas are rare, low-grade, slow-growing primary bone tumors arising from embryonic notochordal remnants [32]. The most common location is the sacrococcygeal region (40%–50%) and the base of the skull (35%–40%) followed by the vertebral bodies (15%–20%) [15, 53]. Distant metastases to the lungs, bone, soft tissues, lymph nodes, liver, and skin have been reported in up to 43% of patients [12, 27, 48].

Treatment of chordomas is described in various studies [2, 4–6, 13, 14, 20, 26, 29, 31, 39, 44, 47, 56, 57]. Its minimal response to chemo- and radiotherapy [6, 13] makes surgery the treatment of choice [14, 47]. Local recurrence rates range from 43% to 85% [2, 4, 5, 20, 26, 29, 31, 56, 57] depending on the surgical margins obtained at initial

Each author certifies that he or she has no commercial associations (eg, consultancies, stock ownership, equity interest, patent/licensing arrangements, etc) that might pose a conflict of interest in connection with the submitted article.

Each author certifies that his or her institution has approved the reporting of these cases, that all investigations were conducted in conformity with ethical principles of research, and that informed consent for participation in the study was obtained.

P. Ruggieri (✉), A. Angelini, G. Ussia, M. Montalti, M. Mercuri
Department of Orthopedics, University of Bologna, Istituto
Ortopedico Rizzoli, Via Pupilli, 1, 40136, Bologna (BO), Italy
e-mail: pietro.ruggieri@ior.it

surgery. Despite the minimal response to radiotherapy, most authors consider it a reasonable alternative for inoperable disease, contaminated surgical margins, or elderly patients [3, 5, 10, 12–14, 30, 39, 42, 43, 50, 51, 57]. Reoperation is associated with increased morbidity because of extensive scarring and difficulty in establishing tissue planes.

Several studies report local recurrence in large series at long-term followup [5, 20, 29]. The surgical margins at initial surgery are the most important factor influencing local recurrence [5, 9, 20, 29, 31, 36, 50]. Resections with wide margins reportedly have a lower local recurrence rate than those with inadequate margins [5, 20].

To confirm previous reports we therefore (1) determined the survival rates in patients with sacral chordoma treated by resection; and (2) determined the effect of surgical margins, previous intralesional surgery, and level of resection on survivorship to local recurrence.

Patients and Methods

We retrospectively reviewed the records of 103 patients with sacral chordoma treated from November 1976 to November 2006. We excluded 47 patients who had palliative surgery (either debulking or intralesional surgery) because their medical conditions contraindicated extensive surgery, chemo-, and/or radiation therapy. This left 56 patients, 32 males and 24 females with a mean age of 55 years (range, 22–76 years). Nine had previous intralesional surgery elsewhere; the remaining 47 patients had primary treatment at the time of diagnosis at our institution. None of these 56 patients had any treatment other than surgery. Excluding three patients who died early postoperatively from complications not associated with the primary tumor, the median followup for the remaining patients from surgery to death or the last followup for the purpose of this study was 9.5 years (range, 3–28 years) (Table 1). All patients gave written informed consent to be included in this study. The study was approved by the Institutional Review Board/Ethics Committee of the authors' institution.

Histologic diagnosis was established by trocar or open biopsy. Three patients had transrectal biopsy elsewhere before admission to our institution; 21 patients had biopsy at our institution, and the remaining 32 patients had biopsy elsewhere and were referred to our institution for further treatment. All histologic slides were reviewed by an experienced pathologist (MA) to confirm diagnosis. Histologic sections were evaluated for the predominance of epithelioid cells versus physaliferous cells, presence and degree of nuclear pleomorphism, spindling, necrosis, and cellularity; dedifferentiation was not observed.

Table 1. Demographics and surgical details of the 56 patients included in this study

Details	Patients
Gender	
Male	32 (57%)
Female	24 (43%)
Age (years)*	44–66
Followup (years)*	9.5; range, 3–28
Previous intralesional surgery elsewhere	9 (34%)
Surgical approach	
Posterior only	19 (34%)
Combined anterior/posterior	34 (61%)
Two-stage anterior/posterior	3 (5%)
Bone resection level	
Above S3	37 (66%)
At or below S3	19 (34%)
Blood loss (L)†	4 (2–22)
Surgical time (hours)†	14 (6–26)
Margins	
Wide	31 (55%)
Wide-contaminated	11 (20%)
Marginal	9 (16%)
Intralesional	5 (9%)

*Values are expressed as mean $\pm$ SD; †values are expressed as mean with range in parentheses; all other values are expressed as number of patients.

Preoperative evaluation included standard radiographs, bone scan, CT scan, and MRI. A CT scan of the chest was performed to assess the presence of lung metastases. All imaging studies were examined by an experienced musculoskeletal radiologist (ER).

The aim of surgery was to obtain a wide resection. Sacral resections were classified according to the level of tumor extension: resections above the S3 vertebra were defined as “proximal” (37 patients [66%]) and those at or below the S3 vertebra were “distal” (19 patients [34%]). All proximal resections were performed by a combined anterior (transperitoneal) and posterior approach; 35 patients had subtotal sacral resection and two total sacral resection. All distal resections were performed by a posterior approach using a longitudinal midline or triradiate incision. The anterior approach allows identification of the pelvic structures, ligation of the internal iliac arteries, and exposure of the sacrum proximal to the tumor. The posterior approach offers the advantages of a single operation,

shorter operative time, and less morbidity but with the risk of possible hemorrhage and injury to pelvic viscera during tumor resection.

The anterior approach was performed in the supine position, occasionally with the assistance of general surgeons (AB, MR). The tumor was approached transperitoneally by a midline laparotomy incision. The posterior parietal peritoneum was opened, the ureters were identified, and the iliac vessels were dissected bilaterally. The middle sacral, lower lumbar segmental, iliolumbar, and internal iliac arteries and veins were ligated to reduce intraoperative bleeding during posterior surgery. Draining veins were ligated or cauterized to improve visibility and lessen blood loss. The sacral foramina were used as landmarks for the anterior sacral osteotomy when free of tumor, and the sacroiliac joint was identified lateral to the lumbar trunk (L4–L5) of the lumbosacral plexus. When feasible, a transverse proximal osteotomy was performed at the desired level of resection. Bone wax was applied to the osteotomy to reduce bleeding and improve visibility. A transabdominal myocutaneous rectus abdominis flap [7] packed within the pelvic cavity was used in six patients. A laparotomy sponge was then placed posterior to the vascular structures and rectum for protection during the posterior resection.

In the posterior approach, dissection was performed through the subcutaneous tissue, fascia, and posterior pelvic muscles as indicated by the preoperative imaging studies. All skin areas, previous biopsy sampling, radiation-induced changes, and scars from previous surgery were excised en bloc. To obtain a wide resection, muscle dissection was performed as distant as possible from tumor margins. When not involved by tumor, the sciatic nerve and sacral nerve roots were identified and preserved. Laminectomy was then performed. In proximal sacral resections, the dural sac was ligated with a double-layer suture. When dissection of the soft tissues was completed, osteotomy of the sacrum and sacroiliac joint was performed, and the tumor was then removed. An osteotomy technique using a threadwire saw (T-saw) [37, 52] for proximal partial lateral sacral osteotomy through the posterior approach was sometimes used. The laparotomy sponges were then removed and hemostasis obtained. Frozen sections of specimens from osteotomy sites and soft tissues were evaluated; in case of histologically proven contamination, further resection was performed. We closed off the posterior wound directly or with a myocutaneous rectus abdominis flap. No patients in this series underwent reconstruction. Surgery ranged from 6 to 24 hours.

Histologically, margins of the tumor specimens were defined according to Enneking [19] as (1) wide in 31 patients (55%); (2) marginal in nine (16%); (3) intralesional in five (9%); (4) and wide-contaminated if intraoperatively the tumor was exposed or its pseudocapsule was seen, but

further tissue was removed finally achieving a wide margin in 11 (19%). In the nine patients who had previous intralesional treatment, the final margins obtained were wide in five, marginal in one, intralesional in one, and wide-contaminated in two.

Postoperative management included bed rest and analgesics and progressive mobilization using a lumbosacral corset as tolerated. The purpose was to mobilize the patients as soon as possible to prevent immobilization-related complications such as deep venous thrombosis and urinary infections. All patients who underwent distal resections were able to walk without external support. Complications were classified as intraoperative, early (within 30 days of surgery), and late (more than 30 days from surgery) (Table 2).

Routine followup evaluation was performed every 3 months for the first 2 years, every 6 months for the next 3 years, and then annually. Each followup evaluation included clinical and neurologic examinations [8] and imaging studies with standard radiographs, CT scan, and MRI; a CT scan of the chest was performed annually. The presence of local recurrence, metastasis, or death was assessed and the patients subdivided as follows: (1) continuously disease-free (if the patient was continuously disease-free to the latest routine followup); (2) disease-free after treatment of local recurrence or metastasis; (3) alive

Table 2. Complications of the 56 patients included in this study

Complications	Patients
Intraoperative	
Death	3 (5%)
Massive hemorrhage*	2 (4%)
Iliac vein injury	2 (4%)
Dura tear	2 (4%)
Rectum perforation	2 (4%)
Stroke	1 (2%)
Early	
Wound dehiscence	37 (70%)
Infection	24 (45%)
Motor deficits (L5, S1; proximal resection)	10 (19%)
Bladder and bowel dysfunction	6 (12%)
Cerebrospinal fluid fistula	3 (6%)
Pelvic hematoma	1 (2%)
Massive muscle necrosis	1 (2%)
Late	
Stress fractures of sacrum (proximal resection)	3 (6%)
Deep venous thrombosis	2 (4%)
Rectus abdominis flap necrosis	1 (2%)

*Acute, life-threatening massive bleeding during dissection or tumor removal with hemodynamic instability that was difficult to control by the anesthesiologists.

with disease, patients with local recurrence or metastasis; and (4) died of disease and died of other disease, patients who died from the tumor or other unrelated causes, respectively. The three patients who died early postoperatively from complications not related to the tumor were excluded.

Continuous variables were expressed as mean $\pm$ SD. Categorical variables were expressed as number of occurrences and percentage of the total patients in a category. Actuarial overall survival and survivorship to local recurrences were analyzed using the Kaplan-Meier survival analysis [40]. Differences in survival were determined with the log-rank test. The effect level of resection, surgical margins, and previous intralesional surgery on survivorship to local recurrence was evaluated using the multivariate Cox regression analysis with stepwise forward procedure [40]. The data were recorded in a Microsoft® Excel® 2003 spreadsheet (Microsoft Inc, Redmond, WA) and analyzed using MedCalc® Software Version 11.1 (MedCalc Software, Mariakerke, Belgium).

Results

Overall survival was 97% at 5 years, 71% at 10 years, and 47% at 15 years (Fig. 1); 34 of the 53 patients (64%) are alive and 19 patients died (Table 3). Survivorship to local recurrence was 65% at 5 years and 52% at 10 years (Fig. 2); 24 of the 53 patients (45%) had local recurrence at a mean time from surgery of 3.7 years (range, 4 months to 12 years). The overall metastasis rate was 30% (16 patients); 11 patients had local recurrence and lung metastases, and five only lung metastases that, in two of these patients, developed after 12 and 13 years.

Fig. 1 Survival was 97% at 5 years, 71% at 10 years, and 47% at 15 years.

Table 3. Oncologic evaluation of the 53 patients* included in this study

Oncologic status	Patients
Continuously disease-free	22 (42%)
Disease-free after treatment of local recurrence or metastases	6 (11%)
Alive with disease	6 (11%)
Dead of disease	17 (32%)
Dead of other disease	2 (4%)

*Three patients who died postoperatively were excluded from oncologic evaluation.

Fig. 2 Survivorship to local recurrence was 65% at 5 years and 52% at 10 years.

Fig. 3 Survivorship to local recurrence was increased ($p = 0.045$) in patients with wide compared with inadequate margins.

Wide surgical margins were associated with increased survivorship to local recurrence ($p = 0.045$) (Fig. 3; Table 4). We found no difference ($p = 0.677$) in survivorship to local recurrence between wide and wide-contaminated surgical margins (Fig. 4). Previous intralesional surgery

Table 4. Incidence of local recurrence related to surgical margins

Treatment	Surgical margins	Number of patients*	Number of patients with local recurrence*
Primary surgery at our institution	Wide	24	8 (33%)
	Wide-contaminated	9	2 (22%)
	Marginal [†]	8	5 (63%)
	Intralesional	3	2 (67%)
Previous intralesional surgery elsewhere	Wide	5	4 (80%)
	Wide-contaminated	2	2 (100%)
	Marginal	1	—
	Intralesional	1	1 (100%)

*Three patients who died postoperatively from complications were excluded; in two of these, the margins were wide and in one intralesional;

[†]five patients had also probably intraoperative contamination.

Fig. 4 Survivorship to local recurrence was not increased ($p = 0.677$) in patients with wide compared with wide-contaminated margins.

Fig. 5 Survivorship to local recurrence was decreased ($p = 0.010$) in patients with previous intralesional surgery compared with patients with primary surgical treatment at our institution.

Fig. 6 No differences ($p = 0.224$) between proximal and distal sacral resection were observed.

was associated with ($p = 0.010$) an increased local recurrence rate (Fig. 5) and it was the independent predictor to local recurrence on Cox regression analysis. We found no association ($p = 0.224$) between a lower level of resection and survivorship to local recurrence (Fig. 6).

Discussion

Surgical resection is considered the treatment of choice for sacral chordoma because it improves local control and disease-free survival [1, 2, 4, 5, 13, 14, 38, 42, 44, 56, 57]. The primary prognostic factor considered is the type of surgical margin attained at initial surgery [5, 9, 20, 29, 31, 36, 50] even when sacrifice of adjacent neurovascular structures is necessary [22–24, 49]. We therefore (1) confirmed the survival reported previously; and (2) determined the effect of surgical margins, previous intralesional surgery, and level of resection on survivorship to local recurrence.

Table 5. Summary of reported cases on the treatment of chordomas of the sacrum

Reference	Patients	Primary/ previous treatment	Local recurrence	Disease- free	10-year survival	Maximum followup (years)	Margins	Treatment per patients
Samson et al. [42]	21	21/0	19%	71%	55%	14	11 wide 3 marginal 7 intralesional	5 surgery only 16 surgery + RT (27–65 Gy)
Thieblemont et al. [51]	10	10/0	84%*	16%*	34%*			9 subtotal surgery + RT (38 Gy) 1 radical surgery + RT (56 Gy)
Ozaki et al. [38]	12	7/5	43% 60%	71% 40%		7.5	6 marginal 5 intralesional	10 surgery only 1 surgery + RT 1 RT only
Cheng et al. [14]	23	23/0	28%*	63%*	65%*			23 surgery ± RT
Yonemoto et al. [56]	13	13/0	46%	46%	29%	14	3 wide 2 marginal 8 intralesional	13 surgery only
Bergh et al. [5]	30	23 primary 7 previous	30% 71%	65% 43%	60% 33%	23	16 wide 3 marginal 4 intralesional 5 wide 1 marginal 1 intralesional	25 surgery only 4 surgery + RT 1 RT only
Baratti et al. [4]	28	20/8	61%	32%	49%	17	11 wide 13 marginal 4 intralesional	28 surgery only
Fuchs et al. [20]	52	52/0	44%	52%	52%	23	21 wide 31 inadequate	30 surgery only 22 surgery + RT (47 Gy)
Park et al. [39]	21	14 primary 7 previous	14% 86%	71% 0%	93% 44%	22 13	3 wide/marginal 11 intralesional 2 wide/marginal 5 intralesional	21 surgery + P/P RT
Hulen et al. [29]	16	16/0	75%	25%	63%	15	9 wide 4 marginal 3 intralesional	7 surgery only 2 surgery + chemotherapy 7 surgery + RT
Hanna et al. [26]	18	18/0	66%	61%		10	10 wide 7 marginal 1 intralesional	18 surgery only
Ahmed [1]	18	18/0	33%	83%	70%	20	7 wide 8 marginal 2 intralesional	2 surgery + chemotherapy + RT 6 surgery + RT 9 surgery only 1 RT only
Ruggieri et al. [current study]	56	47 primary 9 previous	32% 78%	42% 22%	80% 57%	28	35 wide 8 marginal 4 intralesional 7 wide 1 marginal 1 intralesional	56 surgery only

*Rates were recalculated for sacral chordomas only; †previous intralesional surgery performed elsewhere; RT = radiation therapy; P/P RT = proton/photon beam radiation therapy.

We recognize limitations to our study. First, while we report a relatively large number cohort of patients with sacral chordoma, the subgroups are small and we cannot ensure our findings of no differences in wide versus wide-contaminated margins or level of resection do not reflect a Type II error. Second, as with virtually all studies of sacral chordoma, ours is retrospective and some variables are uncontrolled. The relatively long-term followup does allow us to determine overall recurrence rates, however.

Local recurrence rates of 43% to 85% [2, 4, 5, 20, 26, 29, 31, 56, 57] and metastases rates of 5% to 40% at 1 to 10 years [3, 5, 29, 33, 38, 42, 50, 57] have been reported for sacral chordomas (Table 5). In the present series, the overall survival was 97% at 5 years, 71% at 10 years, and 47% at 15 years. At a median followup of 9.5 years (range, 3–28 years), 45% of patients had local recurrence and 30% metastases, two of them long term after diagnosis and treatment.

The surgical margins obtained at initial surgery and previous intralesional surgery are the primary prognostic factors for local recurrence [2, 5, 6, 12, 14, 20, 26, 33, 38, 56, 57]. A surgical resection with wide margins has been associated with a 5% to 17% local recurrence rate compared with 71% to 81% when margins were intralesional or marginal [5, 20]. When gluteal invasion is present, the risk of recurrence is reportedly higher and wider posterior surgical margins are important [56]. To obtain wide margins, usually a combined anteroposterior approach is necessary for proximal chordomas, whereas wide resection of the distal sacrum can be performed through a single posterior approach [2, 15, 16, 42, 49]. With wide resection obtained at primary surgery, survivorship to local recurrence was increased. However, previous intralesional surgery and primary resection with intralesional or marginal margins carries a substantial risk factor for local recurrence.

Intraoperative contamination is a major complication in musculoskeletal tumor surgery [26, 33, 54]. One study reported [33] a local recurrence rate of 28% after en bloc tumor resection compared with a 64% local recurrence rate when the tumor capsule was violated intraoperatively. Another study [26] suggested intraoperative contamination of muscle and/or of the sacroiliac joint increased the probability of recurrence. We classified margins as wide-contaminated when intraoperatively the tumor was exposed or its pseudocapsule was seen, but further tissue was removed finally achieving wide margins, according to Enneking [19]. Although literature [25, 32] describes an increased risk of local recurrence in intraoperative contamination, we found no difference in the survivorship to local recurrence in patients with wide-contaminated compared with wide margins.

Proximal sacral resections are associated with substantial perioperative morbidity, including wound complications

requiring flap closure, mechanical instability requiring spinopelvic reconstruction and fusion, stress fractures, and long-term neurologic deficits [5, 14, 16, 20, 24, 38, 42, 47, 55, 56, 58]. Although there are various solutions, most authors recommend spinopelvic reconstruction after total or high proximal sacrectomy because of instability associated with complete dissociation of the mobile spine with the pelvis [17, 18, 28, 41, 45, 55]. However, other surgeons [11, 34, 46], including us, do not advocate reconstruction of the osseous defect after total sacrectomy because of the risk of major wound complications (especially deep wound infection) and considering the acceptable ambulatory status of patients. Usually, after total sacrectomy, the lumbar spine migrates inferiorly and remains between the ilia [17, 18, 45, 55]. The muscles and scar between the pelvis and spine form a biologic sling eventually stabilizing the spine. Most patients are able to walk with a brace with only slight back or leg pain [25]. Distal sacral chordomas can be resected more easily and with less postoperative disability than proximal tumors [35]. However, in the surgical planning of a distal sacrectomy, preoperative staging of tumor infiltration into the gluteal muscles by MRI is crucial, and vast muscular resection is required for wide resection to reduce the risk of local recurrence [1, 21, 26, 31, 56]. Although we presumed resection at a lower level of the sacrum would relate to survivorship, we found no such difference with our relatively small cohorts.

Primary wide surgical resection is necessary for long-term survival of patients with sacral chordomas. Previous intralesional surgery is associated with higher rate of local recurrence; we therefore believe surgery should be performed in a specialized tumor center with high levels of surgical and oncologic expertise. Intraoperative contamination is a major complication but does not affect survival if the overall margins are wide.

Acknowledgments We thank Dr Marco Alberghini for the histologic evaluation; Dr Eugenio Rimondi for the imaging analysis; Prof Antonio Briccoli and Dr Michele Rocca for surgical assistance of most patients in this series; and Dr Andreas F. Mavrogenis for reviewing our paper. Special thanks to Mrs Alba Balladelli, BA, who reviewed our paper and substantially improved our English.

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, Yildiz Y, Sağlik Y. Management of sacrococcygeal chordomas. *Int Orthop.* 2006;30:514–518.
3. Azzarelli A, Quagliuolo V, Cerasoli S, Zucali R, Bignami P, Mazzaferro V, Dossena G, Gennari L. Chordoma: natural history and treatment results in 33 cases. *J Surg Oncol.* 1988;37:185–191.
4. Baratti D, Gronchi A, Pennacchioli E, Lozza L, Colecchia M, Fiore M, Santinami M. Chordoma: natural history and results in

- 28 patients treated at a single institution. *Ann Surg Oncol*. 2003;10:291–296.
5. Bergh P, Kindblom LG, Gunterberg B, Remotti F, Ryd W, Meis-Kindblom JM. Prognostic factors in chordoma of the sacrum and mobile spine: a study of 39 patients. *Cancer*. 2000; 88:2122–2134.
6. Bethke KP, Neifeld JP, Lawrence WJ. Diagnosis and management of sacrococcygeal chordoma. *J Surg Oncol*. 1991;48:232–238.
7. Biagini R, Ruggieri P, Briccoli A, Fasano D, Montanari FM, Gamberini G, Perin S, Mercuri M. Use of the trans-abdominal rectus abdominis flap in sacrum surgery. *Chir Organi Mov*. 2001;86:1–5.
8. Biagini R, Ruggieri P, Mercuri M, Capanna R, Briccoli A, Perin S, Orsini U, Demitri S, Arlecchini S. Neurologic deficit after resection of the sacrum. *Chir Organi Mov*. 1997;82:357–372.
9. Boriani S, Bandiera S, Biagini R, Bacchini P, Boriani L, Cappuccio M, Chevalley F, Gasbarrini A, Picci P, Weinstein JN. Chordoma of the mobile spine: fifty years of experience. *Spine*. 2006;31:493–503.
10. Breteu N, Demasure M, Favre A, Leloup R, Lescrainier J, Sabattier R. Fast neutron therapy for inoperable or recurrent sacrococcygeal chordoma. *Bull Cancer Radiother*. 1996;83:142S–145S.
11. Capanna R, Briccoli A, Campanacci LC, et al. Benign and malignant tumors of the sacrum. In: Frymore JW, Ducker TB, Hadler NM, Kostuik JP, Weinstein JN, Whitecloud III TS, eds. *The Adult Spine: Principles and Practice*. Philadelphia, PA: Lippincott-Raven Publishers; 1997:2367–2405.
12. Catton C, O'Sullivan B, Bell R, Laperriere N, Cummings B, Fornasier V, Wunder J. Chordoma: long-term follow-up after radical photon irradiation. *Radiother Oncol*. 1996;41:67–72.
13. Chandawarkar RY. Sacrococcygeal chordoma: review of 50 consecutive patients. *World J Surg*. 1996;20:717–719.
14. Cheng EY, Ozerdemoglu RA, Transfeldt EE, Thompson RC. Lumbosacral chordoma: prognostic factors and treatment. *Spine*. 1999;24:1639–1645.
15. Dahlin DC, MacCarthy CS. Chordoma: a study of 59 cases. *Cancer*. 1952;5:1170–1178.
16. Devin C, Chong PY, Holt GE, Feurer I, Gonzalez A, Merchant N, Schwartz HS. Level-adjusted perioperative risk of sacral amputations. *J Surg Oncol*. 2006;94:203–211.
17. Dickey ID, Hugate RR, Fuchs B, Yaszemski MJ, Sim FH. Reconstruction after total sacrectomy. *Clin Orthop Relat Res*. 2005;438:42–50.
18. Doita M, Harada T, Iguchi T, Sumi M, Sha H, Yoshiya S, Kurosaka M. Total sacrectomy and reconstruction for sacral tumors. *Spine*. 2003;28:E296–E301.
19. Enneking WF. A system of staging musculoskeletal neoplasms. *Clin Orthop Relat Res*. 1986;204:9–24.
20. Fuchs B, Dickey ID, Yaszemski MJ, Inwards CY, Sim FH. Operative management of sacral chordoma. *J Bone Joint Surg Am*. 2005;87:2211–2216.
21. Gerber S, Ollivier L, Leclère J, Vanel D, Missenard G, Brisse H, de Pinieux G, Neuenschwander S. Imaging of sacral tumours. *Skeletal Radiol*. 2008;37:277–289.
22. Gunterberg B. Effects of major resection of the sacrum: clinical studies on urogenital and anorectal function and a biomechanical study on pelvic strength. *Acta Orthop Scand Suppl*. 1976;162:1–38.
23. Gunterberg B, Norlen L, Stener B, Sundin T. Neurourologic evaluation after resection of the sacrum. *Invest Urol*. 1975;13: 183–188.
24. Gunterberg B, Romanus B, Stener B. Pelvic strength after major amputation of the sacrum: an experimental study. *Acta Orthop Scand*. 1976;47:635–642.
25. Guo Y, Yadav R. Improving function after total sacrectomy by using a lumbar-sacral corset. *Am J Phys Med Rehabil*. 2002;81: 72–76.
26. Hanna SA, Aston WJ, Briggs TW, Cannon SR, Saifuddin A. Sacral chordoma: can local recurrence after sacrectomy be predicted? *Clin Orthop Relat Res*. 2008;466:2217–2223.
27. Higinbotham NL, Phillips RF, Farr HW, Hustu HO. Chordoma: thirty-five-year study at Memorial Hospital. *Cancer*. 1967;20: 1841–1850.
28. Hüfner T, Kfuri M Jr, Galanski M, Bastian L, Loss M, Pohlemann T, Krettek C. New indications for computer-assisted surgery: tumor resection in the pelvis. *Clin Orthop Relat Res*. 2004;426: 219–225.
29. Hulen CA, Temple HT, Fox WP, Sama AA, Green BA, Eismont FJ. Oncologic and functional outcome following sacrectomy for sacral chordoma. *J Bone Joint Surg Am*. 2006;88:1532–1539.
30. Imai R, Kamada T, Tsuji H, Yanagi T, Baba M, Miyamoto T, Kato S, Kandatsu S, Mizoe J, Tsujii H, Tazaki S; Working Group for Bone, Soft Tissue Sarcomas. Carbon ion radiotherapy for unresectable sacral chordomas. *Clin Cancer Res*. 2004;10: 5741–5746.
31. Ishii K, Chiba K, Watanabe M, Yabe H, Fujimura Y, Toyama Y. Local recurrence after S2–3 sacrectomy in sacral chordoma: report of four cases. *J Neurosurg*. 2002;97(Suppl):98–101.
32. Jemal A, Siegel R, Ward E, Murray T, Xu J, Thun MJ. Cancer statistics, 2007. *CA Cancer J Clin*. 2007;57:43–66.
33. Kaiser T, Pritchard D, Unni KK. Clinicopathologic study of sacrococcygeal chordoma. *Cancer*. 1984;53:2574–2578.
34. Michel A. Total sacrectomy and lower spine resection for giant cell tumor: one case report. *Chir Organi Mov*. 1990;75:117–118.
35. Mindell ER. Risk of sacral amputations. *J Surg Oncol*. 2006;94: 173–174.
36. Osaka S, Kodoh O, Sugita H, Osaka E, Yoshida Y, Ryu J. Clinical significance of a wide excision policy for sacrococcygeal chordoma. *J Cancer Res Clin Oncol*. 2006;132:213–218.
37. Osaka S, Kondoh O, Yoshida Y, Ryu J. Radical excision of malignant sacral tumors using a modified threadwire saw. *J Surg Oncol*. 2006;93:312–317.
38. Ozaki T, Hillmann A, Winkelmann W. Surgical treatment of sacrococcygeal chordoma. *J Surg Oncol*. 1997;64:274–279.
39. Park L, Delaney TF, Liebsch NJ, Hornicek FJ, Goldberg S, Mankin H, Rosenberg AE, Rosenthal DI, Suit HD. Sacral chordomas: impact of high-dose proton/photon-beam radiation therapy combined with or without surgery for primary versus recurrent tumor. *Int J Radiat Oncol Biol Phys*. 2006;65:1514–1521.
40. Petrie A. Statistics in orthopaedic papers. *J Bone Joint Surg Br*. 2006;88:1121–1136.
41. Randall R, Bruckner J, Lloyd C, Pohlman TH, Conrad EU III. Sacral resection and reconstruction for tumors and tumor-like conditions. *Orthopedics*. 2005;28:307–313.
42. Samson IR, Springfield DS, Suit HD, Mankin HJ. Operative treatment of sacrococcygeal chordoma: a review of twenty-one cases. *J Bone Joint Surg Am*. 1993;75:1476–1484.
43. Saxton JP. Chordoma. *Int J Radiat Oncol Biol Phys*. 1981;7: 913–915.
44. Schwab JH, Healey JH, Rose P, Casas-Ganem J, Boland PJ. The surgical management of sacral chordomas. *Spine (Phila Pa 1976)*. 2009;34:2700–2704.
45. Shen FH, Harper M, Foster WC, Marks I, Arlet V. A novel 'four-rod technique' for lumbo-pelvic reconstruction: theory and technical considerations. *Spine*. 2006;31:1395–1401.
46. Simpson AH, Porter A, Davis A, Griffin A, McLeod RS, Bell RS. Cephalad sacral resection with a combined extended ilioinguinal and posterior approach. *J Bone Joint Surg Am*. 1995;77:405–411.
47. Soo MY. Chordoma: review of clinicoradiological features and factors affecting survival. *Australas Radiol*. 2001;45:427–434.
48. Sopta J, Tulic G, Mijucic V, Mamontov P, Mandic N. Solitary lymph node metastasis without local recurrence of primary chordoma. *Eur Spine J*. 2009;18(Suppl 2):191–195.

49. Stener B, Gunterberg B. High amputation of the sacrum for extirpation of tumors: principles and technique. *Spine*. 1978;3: 351–366.
50. Sundaresan N, Huvos AG, Krol G, Lane JM, Brennan M. Surgical treatment of spinal chordomas. *Arch Surg*. 1987;122: 1479–1482.
51. Thieblemont C, Biron P, Rocher F, Bouhour D, Bobin JY, Gérard JP, Blay JY. Prognostic factors in chordoma: role of postoperative radiotherapy. *Eur J Cancer*. 1995;31:2255–2259.
52. Tomita K, Kawahara N: The threadwire saw: a new device for cutting bone. *J Bone Joint Surg Am*. 1996;78:1915–1917.
53. Unni KK. *Dahlin's Bone Tumors: General Aspects and Data on 11,087 Cases*. 5th ed. Philadelphia: Lippincott-Raven; 1996: 291–305.
54. Virkus WW, Marshall D, Enneking WF, Scarborough MT. The effect of contaminated surgical margins revisited. *Clin Orthop Relat Res*. 2002;397:89–94.
55. Wuisman P, Lieshout O, Sugihara S, van Dijk M. Total sacrectomy and reconstruction: oncologic and functional outcome. *Clin Orthop Relat Res*. 2000;381:192–203.
56. Yonemoto T, Tatzaki S, Takenouchi T, Ishii T, Satoh T, Moriya H. The surgical management of sacrococcygeal chordoma. *Cancer*. 1999;85:878–883.
57. York JE, Kaczaraj A, Abi-Said D, Fuller GN, Skibber JM, Janjan NA, Gokaslan ZL. Sacral chordoma: 40-year experience at a major cancer center. *Neurosurgery*. 1999;44:74–79.
58. Zileli M, Hoscokun C, Brastianos P, Sabah D. Surgical treatment of primary sacral tumors: complications associated with sacrectomy. *Neurosurg Focus*. 2003;15:E9.