Long-term Outcomes Following En Bloc Resection for Sacral Tumor: A Retrospective Analysis of 93 Cases

ZHI-YU ZHANG, MD, PHD; CHANG-FENG FU, MD; YING-XIN YANG, MD; LI-QIANG WANG, MD; YAN CUI, MD; YI LIU, MD

abstract

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Sacral tumors are rare. Appropriate surgical resection is crucial to treat the disease while minimizing disease recurrence. We present the results of 93 patients with sacral tumors to analyze the long-term functional and oncological results of patients undergoing en bloc resection. The medical data between January 2003 and July 2010 was retrospectively reviewed. None of the 93 patients died intra- or postoperatively. Patients undergoing intralesional curettage (range, 6500-25,000 mL; mean, 13,500 mL) lost more blood than those patients with wide excision (range, 1000-8100 mL; mean, 3590 mL). Mean follow-up duration from the time of surgery until most recent clinic visit or death was 44.5 months (range, 6-105 months). This study is an educational tool regarding primary sacral tumors and provides evidence on the treatment. It presents results from a large group of patients with sacral tumor. After follow-up, we did not find that sacrificed nerve roots and surgical margins have an impact on the recurrence of the tumor. Patients undergoing intralesional curettage lost more blood than those patients with wide excision. Postoperative bladder/bowel dysfunction was more severe for patients with removal of S1 and S2.

Drs Zhang and Cui are from the Department of Orthopedic Surgery, The 4th Affiliate Hospital of China Medical University, Dr Fu is from the Department of Orthopedic Surgery, The 2nd Affiliate Hospital of Jilin University, and Dr Yang is from the Clinical Department of Soft Tissue Damage, The General Hospital of Shenyang Military Region PLA China, Shenyang, and Drs Wang and Liu are from the Department of Orthopedic Surgery, The China-Japan Friendship Hospital, Beijing, China.

Drs Zhang, Fu, Yang, Wang, Cui, and Liu have no relevant financial relationships to disclose.

Drs Zhang, Fu, and Yang contributed equally to this article.

Correspondence should be addressed to: Zhi-yu Zhang, MD, PhD, Department of Orthopedic Surgery, The 4th Affiliate Hospital of China Medical University, Beiwu Road, Heping District, Shenyang, China (zhangzhiyu_2010@sina.com).

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Sacral tumor is relatively rare and accounts for approximately 1 in 40,000 hospital admissions. The difficulty in diagnosing at the onset is due to a deceptive manifestation. Unfortunately, the tumor is often large when it is definitively diagnosed, which makes surgery challenging. Surgical margin or intralesional curettage does not have a radical effect on sacral malignant tumor, benign tumor, or progressivity tumor (such as giant cell tumor) with a high relapse rate at the local hospital. Radical excision, such as partial or total sacrectomy, has satisfactory treatment effectiveness. However, the intraoperative and postoperative complications including neurological compromise, visceral injury or resection, infection, vascular injury, and severe soft tissue removal threaten the patient’s life and the quality of life.

This article reviews the clinical data on patients with sacral tumor resection to analyze the long-term functional and oncological results of patients undergoing en bloc resection.

**Materials and Methods**

The medical database of our hospital between January 2003 and July 2010 was retrospectively reviewed. Patients were included if the diagnosis was primary sacral tumor, there was no history of prior surgical treatments other than needle biopsy, and there was a minimum of 6 months clinical follow-up. Ninety-three patients with sacral tumor were identified. The data, including patient age, sex, histologic characteristics of primary tumor, neural symptom, surgery technique, level of nerve roots sacrificed, surgical margins, and estimated blood loss were reviewed.

Preoperative evaluation consisted of neurologic examination, sampling biopsy, computed tomography scan, magnetic resonance imaging, abdominal ultrasonography, and 99mTc total body bone scan. All patients underwent preoperative digital subtraction angiography to visualize relationships with internal iliac, mid-sacral and gluteal arteries, and local gel-foam blood vessels embolotherapy was performed.

Surgical margins were grouped as wide, contaminated, or intralesional resections based on the intraoperative findings by the senior surgeon (Z.Z.) and the histopathological examination of the excised tumor specimens. Functional outcomes were determined by analyzing the patients’ bladder and bowel functions at 6-month follow-up.

**Results**

**General Data, Follow-Up Course, and Blood Loss**

The present study consists of 93 consecutive patients who underwent primary en bloc excision of sacral tumor. The study included 48 men and 45 women with ages ranging from 12 to 86 years (mean, 45.1 years). Characteristics of surgical procedures are displayed in Table 1.

All patients were routinely applied tumor-related blood vessel embolism by digital subtraction angiography technique. Twenty-five patients were treated with a posterior-only approach to remove the tumor. Sixty patients underwent a sacrectomy with staged anterior and posterior procedures to achieve en bloc tumor excision. Eight patients were treated with abdomino-anterior approach to remove the tumor. Twenty-six patients of the 67 lesions treated with wide excision, margins were adequate in 52 patients. Surgical margins were intralesional in 5 patients, and contaminated in 10 patients were contaminated. These 15 patients were considered to have inadequate/positive margins. Thus, the overall success rate of achieving our goal of wide en bloc tumor excision was 77.6% in this study.

None of 93 patients died intra- or post-operatively. The intraoperative blood loss ranged from 1000 to 25,000 mL (average, 7250 mL) (Table 2). Twenty-five patients treated with a posterior-only approach lost blood ranging from 1300 to 16,800 mL (mean, 6200 mL), 60 patients with anterior/posterior approach lost 1000 to 22,500 mL (mean, 7400 mL), and 8 patients with abdomino-anterior approach lost 1600 to 25,000 mL (mean, 10,600 mL). In patients undergoing curettage, intraoperative blood loss ranged from 6500 to 25,000 mL (mean, 13,500 mL). In patients with wide excision, intraoperative blood loss ranged from 1000 to 8100 mL (mean, 3590 mL).
Mean follow-up duration from the time of surgery until most recent clinic visit or death was 44.5 months (range, 6-105 months). Ten patients (10.8%) died from tumor metastasis or recurrence in 21 to 101 months, and 24 (25.8%) had local recurrence in 18 to 105 months including malignant neurogenic tumor (2 cases, 2.2%, at 26 and 68 months), malignant fibrous histiocytoma (1 case, 1%, at 97 months), giant cell tumor of bone (7 cases, 7.5%, at 30-96 months), chordoma (13 cases, 14.0%, at 15-70 months), and benign neurogenic tumor (1 case, 1%, at 60 months) who all recovered after complete resection.

Our analysis found improvement of disease-free survival in patients with wide en bloc tumor margins compared to those with contaminated or intralesional margins. Age, sex, and nerve roots sacrificed had no statistically significant impact on the recurrence (Table 3). At the time of analysis, local recurrence was statistically significantly affected by a wide surgical margins $(P=0)$. Of the 52 patients with wide margins 45 (86.5%) had no evidence of disease, and they had a mean disease-free survival of 40.5 months (range, 6-93 months). However, 60.0% (6/10) of those patients with contaminated margins were alive with disease who had mean disease-free survival of 45.3 months (range, 21-68 months), and the other 4 patients had a mean disease-free survival of 22.3 months (range, 6-40 months). One of 5 patients with intralesional margins had recurrence at 35 months, and the other 4 patients were disease free until the last follow-up (average, 30.3 months; range, 12-57 months). Among the 26 patients with intralesional excision, 6 patients had no evidence of disease (average, 38.3 months; range, 19-95 months), 8 died from disease with a mean survival of 52.5 months (range, 21-101 months), and 12 had recurrence at a mean disease-free survival of 40.7 months (range, 16-75 months).

Regarding histopathological diagnosis, 28 of the tumors were diagnosed as giant cell tumor of bone (30.1%), 40 as chordoma (43.0%), 14 as malignant neurogenic tumor (15%), 4 as benign neurogenic tumor (4.3%), 4 as hemangiopericytoma (4.3%), 3 as malignant fibrous histiocytoma (3.2%), and 4 as malignant teratoma (4.3%).

Pre- and Postoperative Functional Outcomes

Postoperative functional outcomes in this study were determined by assessing the patients' bladder and bowel functions 6 months postoperatively. All 32 patients who presented with loss of bladder and bowel control had no control after surgery. Of the 61 patients who had intact bladder and bowel control preoperatively, all 4 patients with S1, and below nerve roots excised lost normal control of urinary and bowel continence, and 4 of 18 patients with S2 and below roots excised, 22 of 31 patients retained normal urinary and bowel continence, and all 8 patients S4 roots excised had normal urinary and bowel function (Table 4).

However, the remaining 63.4% of the patients had either partial or complete loss of bladder or bowel functions. The postsurgical bladder and bowel function was correlated with intraoperative nerve root excised (Table 4).

Among the 61 patients who had intact bladder and bowel control preoperatively, all 4 patients with S1, and below nerve roots excised lost normal control of urinary and bowel continence, and 4 of 18 patients with S2 and below roots excised, 22 of 31 patients retained normal urinary and bowel continence, and 22 of 35 patients retained S3 and below roots excised, and all 8 patients S4 roots excised had normal urinary and bowel function (Table 4).

DISCUSSION

Most of original clinical situations of sacral tumor were pain in the sacrococcygeal region, sometimes accompanied by pseudo or true root radiating pain, and subsequent sense-movement disorder of
Several factors influence local recurrence after the removal of these tumors. Among them, tumor location is the most important factor in recurrence, as statistically significant differences were found between sacral tumor located at or below S3 and located above S3. However, we did not find these differences. Among 50 patients with tumors located above S3, 18 patients had recurrence, and 17 of 43 patients with tumors at S3 and below had recurrence. In addition, age, sex, and surgical margins had no statistically significant impact on the recurrence.

The postoperative local recurrence rate is directly related to the scope and degree of the resection. In our study, 11 of the 37 patients had a wide margin, 11 of which experienced recurrence (29.7%); 21 patients had a perilesional resection, 9 of which experienced recurrence (42.8%); 35 patients had a contaminated excisions, 15 of which experienced recurrence (42.8%). Although we applied tumor-related blood vessel embolism using digital subtraction angiography technique, we did not find a lower recurrence rate for radical resections.

Sectioning of the S1, S2, and S3 nerve roots may result in bladder and bowel disability. Patients in whom all sacral nerve roots have been sacrificed also experienced complete bladder and bowel dysfunction. Postoperative functional outcomes including bladder and bowel functions were assessed 6 months postoperatively. We found that 27 of 61 patients with intact bladder and bowel control preoperatively experienced postoperative bowel and/or bladder dysfunction at 6 months. Postoperative bladder/bowel dysfunction was more severe for patients with removal of S1 and S2 (only 4 had normal urinary or bowel function).

Within the spine, chordoma is the most common primary tumor of the sacrum, and giant cell tumor is the second most common. Randall et al found chordoma was the most common primary tumor (6 patients). Other diagnoses included chondrosarcomas (3), osteosarcoma (3), Ewing’s sarcoma (3), malignant peripheral nerve sheath tumor (2), metastatic adenocarcinomas (2), fibrosarcoma (1), hemangiopericytoma (1), monophasic synovial sarcoma (1), lymphoma (1), ependymoma (1), and hibernoma (1). In our study, most patients were diagnosed with giant cell tumor and chordoma with histopathological diagnosis. Twenty-eight patients had giant cell tumor of bone (30.1%), and 40 chordoma (43.0%). Other diagnoses included malignant neurogenic tumor (14), benign neurogenic tumor (4), hemangiopericytoma (4), malignant fibrous histiocytoma (3), and malignant teratoma (4).

Among the 26 patients with intralesional excision, only 6 patients had no evidence of disease, 76.9% suffered from recurrence, and 8 patients died from disease. The recurrence rate was higher than Turcotte et al reported, which was 33%. This may be due to the fact that there were various type of tumors in these 26 patients: consisted of 15 chordoma (11 cases of recurrence), 5 giant cell tumor (4 cases of recurrence), 1 hemangioendothelioma (died from disease), 2 malignant fibrous histiocytoma (1 patient died from disease), and 3 malignant neurogenic tumor (2 cases of recurrence).

Our study has several limitations. It is a retrospective review. Although we have collected data of 93 patients, a relative large number, the pool of patients from several centers represents with varying malignant and benign pathologies. The duration of follow-up is relatively short (range, 6-105 months), which might affect the conclusions drawn. In addition, we could not review enough data of sexual function and motor function.

Despite these shortcomings, this study can help us to understand primary sacral tumors and provide some evidence on the treatment. It presents results from a large group of patients with sacral tumor. After follow-up, sacrificed nerve roots and surgical margins were not found to have an impact on the tumor recurrence. Patients undergoing intralesional curettage lost...
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REFERENCES