Surgical Consideration for Patients with Repeatedly Recurrent Spinal Chordoma: A Systematic Analysis of Prognostic Factors and Quality of Life

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Abstract

Background: A retrospective study of patients with repeatedly recurrent spinal chordoma (RRSC) was performed. The objective of this study was to evaluate the independent prognostic factors for RRSC improving life expectancy and discuss the most appropriate treatment modality. Methods: Medical data and follow-up record of patients who were diagnosed as RRSC and were surgically treated in Changzheng hospital between July 2010 and September 2017 were reviewed systematically. Univariate and multivariate analysis were performed to identify possible independent prognostic factors for patients with RRSC. Recurrence-free survival (RFS) and overall survival (OS) were estimated by Kaplan-Meier method. Factors with $P < 0.1$ were subjected to multivariate analysis by Cox regression analysis. $P < 0.05$ was considered statistically significant. Results: Sixty-five consecutive patients with RRSC were included in this study. Local recurrence was detected in thirty-three patients, while death was occurred in twenty-one patients. The mean follow-up period was 34.3 months. The statistical results revealed that number of recurrence (NOR), surgical method, and surgical margin were independent prognostic factors for RFS and preoperative Frankel score (PFS) was favorable prognostic factor for OS. Moreover, subtype analysis suggested that treatment method could make significant difference for prognosis of patients with RRSC. Conclusion: Less NOR, total resection, and wide surgical margin could significantly reduce the risk of local recurrence of RRSC. PFS of A-C was adverse prognostic factor for OS. Total en bloc resection or total piecemeal resection with postoperative radiotherapy is recommended as ideal treatment method for prolonging life expectancy.

Background

Chordoma is one of relatively uncommon low malignant bone tumors, which is originated remnants of the embryonal notochord and occurs along spinal axis from skull base to sacrococcygeal spine [1, 2]. The incidence rate of chordoma is approximately 0.8 per 100000, with clear male predominance and a peak occurrence in late fifties in age [3, 4]. The distribution of spinal chordoma is not average, mostly located in sacrococcygeal region followed by skull base then the mobile spine [5]. In clinic, radical surgical resection especially en bloc resection is recommended as standard treatment modality of spinal chordoma, and adjuvant radiotherapy was combined with inadequate margins or planned incomplete resection [6].

Despite its low-grade malignance and slow growing pattern, spinal chordoma exhibits poor prognosis because of aggressively local invasion with high rate of recurrence, but appears a low to late tendency for distant metastasis. Published literature indicated that the 5-year overall survival (OS) rate and 5-year recurrence-free survival (RFS) rate of primary spinal chordoma ranged from 78–82% and 47–72%, respectively [7]. However, it is consistent theme that the outcome of recurrent spinal chordoma is uniformly worse than that of primary tumor. Compared to primary cases, the 5-year OS rate and RFS rate were a range of 47–78% and 36–50%, respectively [8, 9]. Owing to its rarity, limited information regarding to clinical experience and prognostic analysis of spinal chordoma has been reported, but most of published information only focused on prognosis of primary spinal chordoma, with less attention on recurrent cases especially repeatedly recurrent cases. Tamir Ailon et al [10] systematically reviewed main

published articles about recurrent spinal chordoma and obtained guidance in regard to the management of recurrent cases. Wide en bloc resection should be achieved if technically feasible with appropriate cases, and high-dose radiation therapy was suggested to administer postoperatively to reduce the risk of local recurrence. Nevertheless, repeatedly recurrent spinal chordoma (RRSC) was extremely excellent challenge for surgeon with unclear surgical margins and complex anatomic structure. Thus, there was no published information about the surgical consideration and prognostic analysis of RRSC.

Therefore, it was necessary to perform a systematic prognostic analysis of a large case series about RRSC. The objective of this study was to evaluate the independent prognostic factor improving life expectancy and to discuss the most appropriate treatment modality.

Methods

Patients

A retrospective study was performed of 65 patients with RRSC who was surgically treated between July 2010 and September 2017 in our spinal tumor center. All patients were diagnosed as spinal chordoma according to clinical manifestation, radiological results, and postoperative pathological outcome by two different pathologists, and they have experienced at least two-time recurrence. RRSC was defined as spinal chordoma that experienced twice or more than twice recurrence. Permission was obtained from the Ethic Committee of Changzheng hospital before this retrospective study, informed consent was approved by patients or their legal guardians. A typical patient's material was shown in Fig. 1.

Medical data of all patients was obtained from the database of our hospital, and we reviewed clinical and surgical records, radiological images, and pathological outcomes. Patients' preoperative neurological status was evaluated by preoperative Frankel score [11]. Tumor classification was determined using Tomita classification and Ennecking stage according to CT images and MRI findings [12, 13]. Chordoma was grouped into three categories including classical, chondroid, and dedifferentiated subtypes, according to postoperative pathological appearance [14]. The first surgical opportunity and treatment method are extremely crucial for patients with spinal chordoma, which is related to patients' prognosis. Hence, first surgical institution regarded as one possible prognostic factor was included in the study. Surgical margins were divided into wide margin and narrow margin including marginal or intralesional margins [15]. The study focused on the recurrence and death status for patients with RRSC, thus recurrence-free survival (RFS) and overall survival (OS) were the end point events. Event times were defined as the interval from the date of surgery to local recurrence, death, or until September 2017 for living patients. All patients were followed up on an outpatient basis at 3-month intervals for the first 6 months, then at 6-mo intervals for the next 2 years, and annually.

Statistical method

Quantitative data are described by mean (median, range), and qualitative data are described as counts and percentages. The RFS rate and OS rate were estimated by Kaplan-Meier method. The univariate and

multivariate analysis were used to identify various clinical variables for prognostic prediction. Clinical experience and statistical analysis were used to decide whether continuous variables should be categorized. The log-rank test was performed for univariate analysis, and all factors with P value ≤ 0.1 in univariate analysis were extracted into multivariate analysis by Cox proportional hazard analysis. P values ≤ 0.05 were considered statistically significant. All statistical calculations were performed using SPSS Statistics, version 21.0 (IBM, Armonk, New York).

Results

Patients' baseline information

The characteristics of all patients were described in Table 1. The population comprised of 44 males and 21 females with a mean age of 54.2 years (median, 56 y; range, 16–78 y). In this series, two-time recurrence occurred in 32 patients, and more than two-time recurrence occurred in the remaining patients. Lesions were detected in cervical spine (19), thoracic spine (2), lumber spine (6), and sacrum (38), respectively. 52 cases were diagnosed with classical chordoma, 5 with chondroid variants, and 8 with the differentiated subtype. All patients experienced surgical treatment including subtotal resection in 27 cases, total piecemeal resection in 34 cases, and en bloc resection in 4 cases. Of 65 patients, 32 patients were performed first surgical treatment in our hospital, and the remaining 33 patients were firstly surgically treated in other hospitals. 28 surgical margins were wide, and the other were narrow.

Table 1

Patients' baseline information and univariate analysis of prognostic factors affecting recurrence-free survival and overall survival.

Factors	Number	Recurrence-free survival		Overall survival	
			p		p
		median time (months)	value	median time (months)	value
Gender					
Male/female	44/21	24 vs 27	0.676	31 vs 34	0.827
Age					
< 50 y/ >= 50 y	16/49	24 vs 27	0.968	26 vs 33	0.612
NOR					
2 times/ > 2 times	32/23	38 vs 17	< 0.001*	32 vs 29	0.024*
DOS					
< 6 mon/ >= 6 mon	46/19	26 vs 27	0.596	30.5 vs 34	0.806
PFS					
A-C/D-E	36/29	28.5 vs 24	0.537	34 vs 30	0.033*
Tumor location					
Cervical/thoracic/lumber/sacral	19/2/6/38	18 vs 24 vs 24.5 vs 28.5	0.174	29 vs 34.5 vs 34.5 vs 33	0.325
Involved segment					
Monosegment/multisegment	3/62	38 vs 26	0.765	38 vs 33	0.655
Ennecking grading					
I-II/III	30/35	29.5 vs 24	0.857	33 vs 31	0.712
Tomita classification					
I-III/IV-VII	31/34	29 vs 21.5	0.064*	32 vs 30.5	0.336

Factors	Number	Recurrence-free survival		Overall survival	
Histological subtype					
Classical/chondroid/dedifferential	52/5/8	28.5 vs 19 vs 18.5	0.006*	31.5 vs 30 vs 29	0.050*
Tumor size					
<= 6 cm/ > 6 cm	25/40	30 vs 24.5	0.386	31 vs 30	0.302
Surgical pathway					
Anterior/posterior/combined	9/48/8	15 vs 26.5 vs 29	0.070*	26 vs 31 vs 34.5	0.196
Surgical method					
Subtotal/total piecemeal/ total en-bloc	27/34/4	16 vs 28.5 vs 40	0.003*	27 vs 34.5 vs 40	0.024*
Intraoperative bleeding					
<= 2000 mL/ > 2000 mL	49/16	27 vs 24	0.962	31 vs 27	0.284
Intraoperative chemotherapy					
Yes/no	60/5	26.5 vs 12	0.523	31.5 vs 25	0.716
Postoperative radiotherapy					
Yes/no	29/36	28.5 vs 24	0.588	35.5 vs 29	0.418
FSI					
Our hospital/others	32/33	29 vs 24	0.052*	31.5 vs 30	0.034*
Surgical margin					
Wide/narrow	28/37	22 vs 29	0.004*	31 vs 25.5	0.103
Distant metastasis					
Yes/no	10/55	29.5 vs 27	0.135	31 vs 43.5	0.035*
Recurrence					

Factors	Number	Recurrence-free survival	Overall survival	
Yes/no	33/32		25 vs 35.5	0.012*
* Indicates statistical significance (p < 0.10)				
NOR: number of surgery; DOS: duration of symptom; PFS: preoperative Frankel score; FSI: first operative institution				

The mean follow-up period was 34.3 months (median, 33 months; range, 9–86 months). Recurrence was detected in 33 patients after surgical treatment, and 21 patients died during follow-up period. The mean time from surgery to recurrence was 27.0 months (median, 13 months; range, 3–53 months), while follow-up for the dead patients was 32.9 months (median, 20 months; range, 9–65 months).

Univariate analysis of prognostic factors for recurrence-free survival and overall survival

The univariate analysis of prognostic factors for RFS and OS was shown in Table 1. The RFS rate after surgery for patients with RRSC was 49.2%, and the mean RFS was 28.6 months (median, 26 months; range, 3–86 months). In our series, patients experienced two-time recurrence had less risk of recurrence than those experienced more than two-time recurrence (P < 0.001). Besides, patients with tumor classified as Tomita I-III had longer RFS than those with tumor classified as Tomita IV-VII (P = 0.064). The difference was found in cases with classical, chondroidal, and dedifferentiated chordomas (P = 0.006). In addition, patients who were performed different surgical pathway had significantly different RFS (P = 0.070). The RFS rate was obviously different in patients undertaken three kinds of resection methods (P = 0.003). Compared to patients who was firstly treated in other hospital, those patients who chose our spinal tumor center as first surgical institution had significantly high RFS rate (P = 0.052). Moreover, wide surgical margin can significantly improve patients' RFS (P = 0.004). There was no significant difference in other factors.

The OS rate for those patients was 67.7%, and the mean OS was 34.3 months (median, 31 months; range, 9–86 months). Univariate analysis indicated that NOR (P = 0.024), PFS (P = 0.033), histological subtype (P = 0.050), surgical method (P = 0.024), FSI (P = 0.034), distant metastasis (P = 0.035), and recurrence (P = 0.012) were potential independent prognostic factors affecting OS, respectively.

Multivariate analysis of prognostic factors for recurrence-free survival and overall survival

Potential prognostic factors extracted by univariate analysis were subjected into Cox proportional hazard analysis (Table 2, 3). The risk of recurrence of patients with NOR more than two times was significantly higher than that of patients with NOR of two times (The hazard ratio [HR] was 0.417, P = 0.040). The risk of recurrence was significantly decreased in patients who underwent total resection including total

piecemeal resection and en bloc resection (The hazard ratio [HR] was 0.416, P = 0.018). Narrow surgical margin could increase the risk of recurrence (The hazard ratio [HR] was 2.45, P = 0.027). In addition, Patients with PFS classified as A-C had significantly shorter OS than those with PFS classified as D-E (The hazard ratio [HR] was 0.157, P = 0.005). The Kaplan-Meier curves of RFS for NOR, surgical method, and surgical margin were shown in Fig. 2. The Kaplan-Meier curve of OS for PFS was shown in Fig. 3.

Table 2
Multivariate analysis of prognostic factors affecting recurrence-free survival

Factors	B	Exp(B)	95%CI	p value
NOR	-0.875	0.417	0.181–0.959	0.040*
Tomita classification	0.496	1.642	0.727–3.708	0.233
Histological subtype	0.264	1.302	0.774–2.192	0.320
Surgical pathway	-0.585	0.557	0.270–1.148	0.113
Surgical method	-0.877	0.416	0.201–0.862	0.018*
FSI	0.131	1.139	0.494–2.630	0.760
Surgical margin	0.896	2.45	1.105–5.432	0.027*
*Indicates statistical significance (p < 0.05)				
NOR: number of recurrence; FSI: first operative institution				

Table 3
Multivariate analysis of prognostic factors affecting overall survival

Factors	B	Exp(B)	95%CI	p value
NOR	0.689	1.991	0.651–6.091	0.227
PFS	-1.853	0.157	0.043–0.567	0.005*
Histological subtype	-0.030	0.971	0.491–1.921	0.932
Surgical method	0.336	1.399	0.446–4.386	0.564
FSI	-0.105	0.901	0.303–2.680	0.851
Metastasis	0.336	1.399	0.470–4.165	0.547
Recurrence	-13.873	0.000	0.000-1.744E + 082	0.894
*Indicates statistical significance (p < 0.05)				
NOR: number of recurrence; PFS: preoperative Frankel score; FSI: first operative institution				

Treatment method and outcome for patients with RRSC

Surgical excision and postoperative radiotherapy (RT) were performed in patients with RRSC. Subtype analysis was described in Table 4. Subtotal resection, subtotal resection + RT, total piecemeal resection, total piecemeal resection + RT, and En bloc resection were used in 15, 12, 16, 18, and 4 cases, respectively. Statistical analysis revealed that treatment method could make a significant difference in RFS (P < 0.001) and OS (P = 0.006), and patients undertaken total piecemeal resection and en bloc resection had longer RFS and OS. The Kaplan-Meier curves of RFS as well as OS for treatment method were shown in Fig. 4.

Table 4
Treatment method and outcome for patients with RRSC.

Treatment method	Number	Recurrence-free survival (RFS)			Overall survival (OS)		
		Number	%	p value	Number	%	p value
Subtotal resection	15	2	13.3	< 0.001*	5	33.3	0.006*
Subtotal resection + RT	12	5	41.7		9	75.0	
Total piecemeal resection	16	7	43.8		11	68.8	
Total piecemeal resection + RT	18	14	77.8		15	83.3	
En bloc resection	4	4	100		4	100	
Indicates statistical significance (p < 0.05)							
RT: postoperative radiotherapy							

Discussion

With the spinal complex anatomic structure and high potential of recurrence, the management of recurrent spinal chordoma was extremely big challenge for surgeons [16]. As for RRSC, how to decrease the risk of recurrence and prolong the OS was a tough issue due to its locally aggressive behavior and its resistance to adjuvant therapies [17, 18]. Notably, it is the first time to take NOR and FSI into consideration of prognosis of patients with RRSC. Moreover, there was no published information regarding prognosis of RRSC. In our study, univariate and multivariate analysis revealed that RFS was significantly associated with NOR, surgical method, and surgical margin, and OS was only related to PFS. Subtype analysis indicated that en bloc resection as well as total piecemeal resection with postoperative radiotherapy could beneficially improve RFS and OS. Besides, our data confirmed the traits of main age distribution in late fifties and clear man predominance once again [19, 20].

In our series, NOR of two times was detected in 32 patients, and NOR more than two times was found in the remaining cases. As for RRSC, scar tissue hyperplasia is extremely obvious in the operation area, which causes the unclear tumor boundary, roughly complete tumor resection, tumor residual, and operating contamination. Simultaneously, recurrent tumor may exacerbate the neurologic defects, reduce the quality of life, and worse life expectancy. Thus, patients with more NOR have more re-recurrent possibility. Stacchiotti et al had reported that 5- and 10-year RFS rates for patients after first relapse were 47% and 31%, and 5- and 10-year OS rates for patients after first relapse were 50% and 26% [7]. Xie et al described that 5- and 10-year OS rates for 30 patients with local recurrent chordoma were 56% and 19% at first recurrence [8]. Our statistical results revealed that RFS rate was 49.2% and OS rate was 67.7%, which was consistent with published information [21, 22]. The first surgical opportunity is vital for surgeons and

patients, so radical surgical treatment as well as adjuvant therapy was necessary in order to decrease the risk of recurrence.

Given its resistance to radiotherapy and unresponsiveness to chemotherapy, surgical treatment is still the mainstay of treatment modalities for patients with RRSC. In recent published information, it is strongly recommended that total resection including en bloc resection as well as total piecemeal resection could significantly improve the RFS and OS for patients with spinal chordoma. If possible, en bloc resection for spinal chordoma is supposed to achieve [23, 24]. As for RRSC, lesions were hardly excised in en bloc manner because of complex neurovascular structure, unclear scar hyperplasia, and high technically surgical demanding. Thus, total piecemeal resection was performed in these situations, which may cause insufficient surgical margin and tumor contamination. In the subtype analysis, the combination between total piecemeal resection and postoperative radiotherapy could significantly improve the RFS and OS of patients with RRSC compared to only total piecemeal resection. Furthermore, the combination between subtotal resection and postoperative radiotherapy was considered as a useful treatment method that can achieve a comparable outcome with wide surgical resection in the management of chordoma [25].

However, our subtype analysis confirmed that subtotal resection combined with postoperative radiotherapy cannot effectively prolong RFS and OS. Taken together, wide en bloc resection is recommended as optimal treatment method if technically feasible with acceptable morbidity, and total piecemeal resection combined with postoperative radiotherapy is another treatment choice.

Surgical margins were measured on the histological slides in centimeters from the resection surface to the nearest viable tumor, which were grouped into narrow surgical margin and wide surgical margin. Meanwhile, narrow surgical comprised of marginal margin and intralesional margin. The principles of surgical management of RRSC was based on Weinstein-BorianiBiagini (WBB) staging system and Enneking system. Within the thecal sac, the spinal cord and cauda equina are encompassed by osseous structures and, with rare exceptions, cannot be resected to achieve adequate margins. Thus, wide resection and radical resection were unique challenge [10, 26]. Bergh et al had reported that 10 recurrent patients all underwent intralesional resection before referral to the authors' center, and patients experienced wide resection had longer RFS and OS than those with marginal or intralesional resection [27]. Similar results about prognostic significance of surgical margin were found in the literature of Boriani et al and Kayani B [4, 28]. However, our statistical results revealed that wide surgical margin could significantly prolong the RFS of patients, but have no meaningful influence on OS.

In clinical practice, Frankel score was widely used to evaluate injury of spinal cord and physical condition, thus we performed preoperative Frankel score to assess patients' neurological status [11, 28]. Recurrence of tumor could cause tumor rapid proliferation, distant metastasis, and worsen neurological defect, which was related to patients' life expectancy. As for RRSC, radical treatment usually need to sacrifice important neurovascular structure including vertebral artery, nerve root, and even spinal cord, which leads to neurological dysfunction and reduced quality of life. Multivariate analysis indicated that PFS was only significantly independent prognostic factor affecting OS. Though the distant metastasis rate is low in

spinal chordoma, distant metastasis of RRSC cannot be ignored. There were 10 patients detected distant metastasis in our research, and 8 patients experienced next recurrence and died during follow up. York et al had reported that 5-year survival rate for spinal chordoma patients without distant metastasis was 86% compared with 40% for those in metastatic stage [29]. However, distant metastasis had no prognostic significance in our study.

To our knowledge, our study is the first time to evaluate prognostic factors for RRSC, and this is the largest case series for recurrent spinal chordoma. Nevertheless, there are some limitation in our study. Firstly, this is a retrospective study. Secondly, the duration of follow-up is not long enough, which makes OS appear higher than it actually was. Thirdly, we only focus on surgical patients, and neglect patients who did not accept surgery.

Conclusion

Less NOR, total resection including total piecemeal resection and total en bloc resection, and wide surgical margin were favorite prognostic factors for RFS of patients with RRSC. In addition, PFS was only independent prognostic factor predicting OS. En bloc resection was optimal treatment if possible, and total piecemeal resection combined with postoperative radiotherapy is another treatment modality in prolonging the life expectancy of these patients.

Abbreviations

OS: overall survival; RFS: recurrence-free survival; RRSC: repeatedly recurrent spinal chordoma; NOR: number of surgery; DOS: duration of symptom; PFS: preoperative Frankel score; FSI: first operative institution; HR: hazard radio; RT: postoperative radiotherapy; WBB: Weinstein-BorianiBiagini

Declarations

Ethics approval and consent to participate

All procedure involving human participants performed in studies were approved by the ethics committee of Changzheng hospital, and informed consents were obtained from all patients or their legal guardians.

Consent for publication

Patients or their legal guardians know and approve the publication.

Availability of data and material

Not applicable

Competing interests

No conflict of interest exits in the submission of this manuscript, and manuscript is approved by all authors for publication. I would like to declare on behalf of my co-authors that the work described was original research that has not been published previously, and not under consideration for publication elsewhere, in whole or in part.

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Authors' Contributions

Jianru Xiao offered the research direction regarding the prognostic factors for repeatedly recurrent spinal chordoma. Quan Huang and Dongjie Jiang designed the study and gave us several meaningful suggestions. Bo Li, Lin Li, and Yuduo Xu reviewed and collected the medical records. Yue Zhang and Xiaolin Li analyzed and interpreted the patient data. Kehan Xu was a contributor in writing the manuscript. All authors read and approved the final manuscript.

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Figures

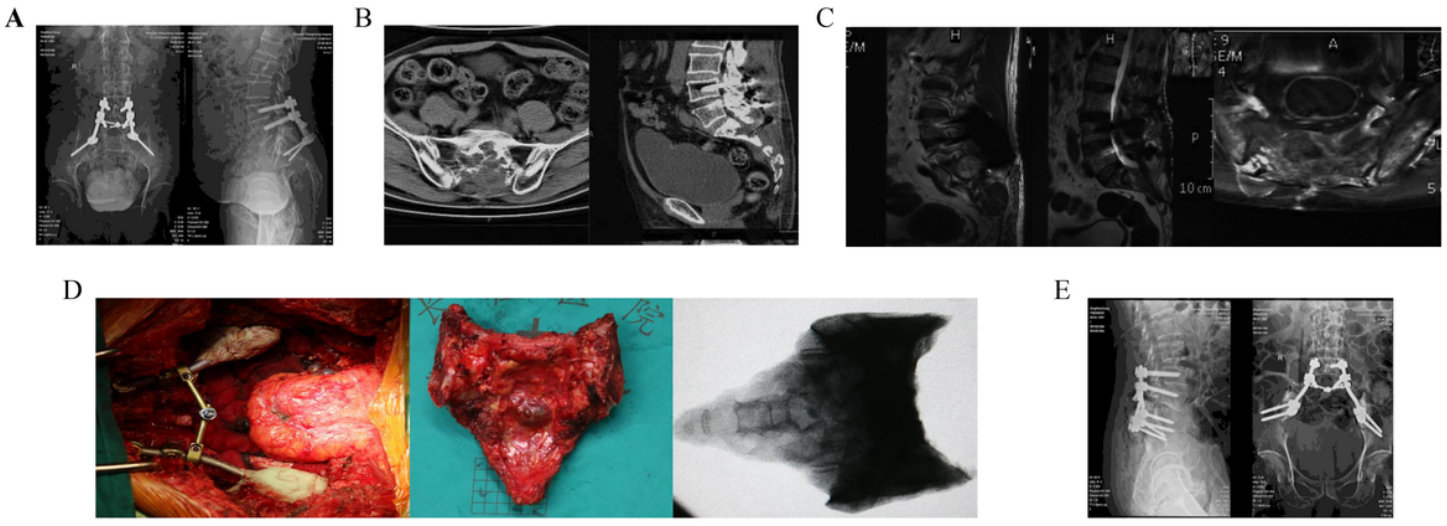

Figure 1

A typical case underwent the removal of tumor by total en bloc resection in our center and was diagnosed as repeatedly recurrent spinal chordoma. (A) Preoperative X-rays of anteroposterior and lateral spine demonstrated metal fixed shadow, and osseous destruction in sacral spine. (B) Preoperative CT showed osteolytic destruction in sacral spine and its posterior elements, paravertebral soft tissue mass, and compression of spinal cord. CT=computed tomography. (C) Preoperative magnetic resonance imaging (MRI) indicated low-intensity signal on T1-weighted image, high-intensity signal on T2-weighted image, and uneven enhancement in enhanced image. (D) The specimen of the tumor by total en bloc resection. (E) The postoperative X-rays showed the sacral spines were removed and replaced by solid internal-fixation.

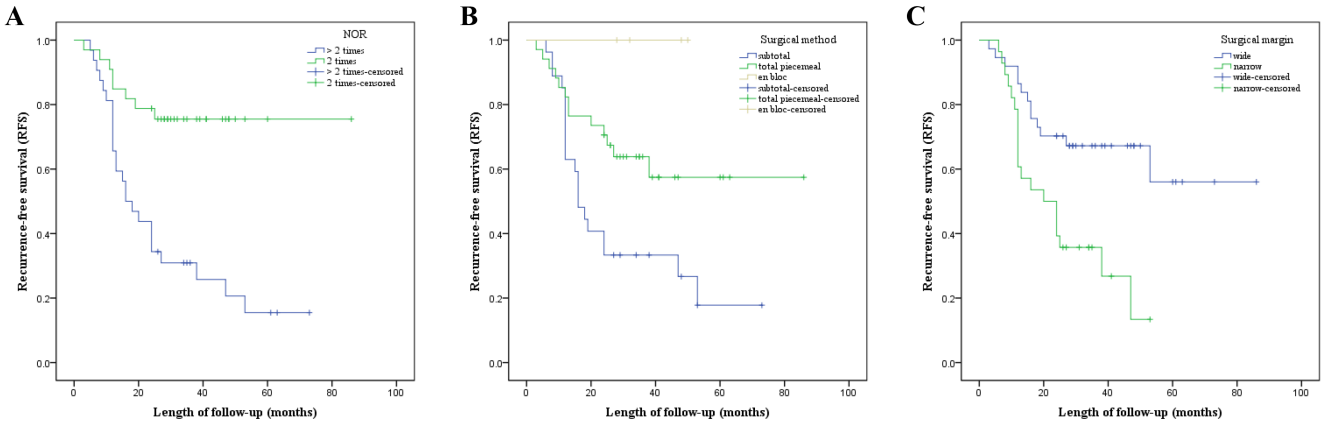

Figure 2

(A) Kaplan-Meier curves of recurrence-free survival for NOR. (B) Kaplan-Meier curves of recurrence-free survival for surgical method. (C) Kaplan-Meier curves of recurrence-free survival for surgical margin.

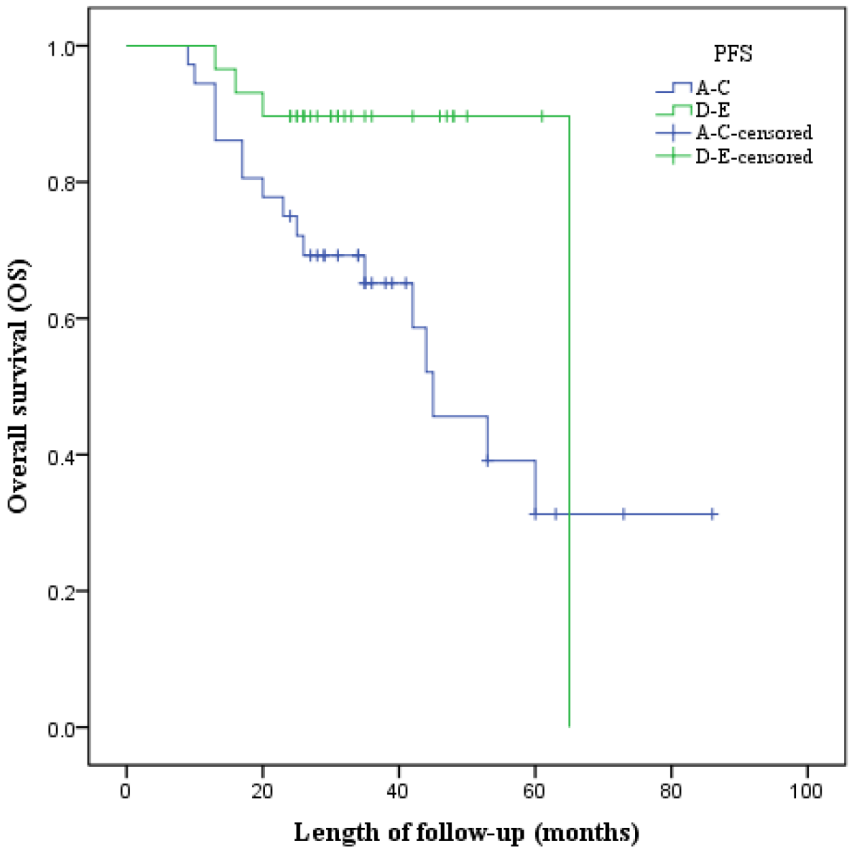

Figure 3

Kaplan-Meier curves of overall survival for preoperative Frankel score.

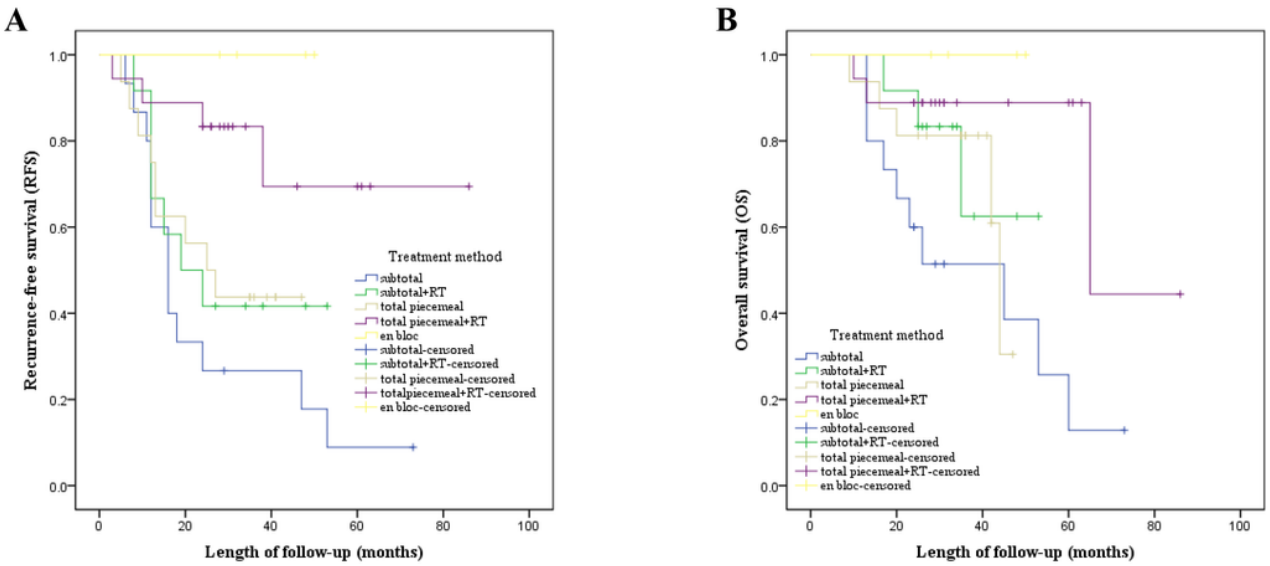

Figure 4

(A) Kaplan-Meier curves of recurrence-free survival for treatment method. (B) Kaplan-Meier curves of overall survival for treatment method.

Supplementary Files

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