

# Primary Osseous Chondrosarcoma in the Lumbar Spine: Case Report and Literature Review With Analysis

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## Case report

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# Abstract

## Background

Primary spinal chondrosarcoma (CS) is relatively rare, with a reported prevalence rate of less than 10% of all CS. En bloc surgery and resection without entering the tumor capsule typically provides a better prognosis. However, the approaches to such tumor lesions vary. Here, we present a case of CS in the lumbar spine treated with two-stage (anterior and posterior approach) en bloc surgery, along with an analysis and comparison of the characteristics and prognosis of one-stage versus two-stage surgery patients in previous studies.

## Case description

A 30-year-old male with an L3 vertebral body CS presented with back pain and lower limb weakness. Lumbar spine MRI showed an L3 vertebral body tumor with cord and root compression. A two-stage surgery including posterior total laminectomy, transpedicular screw fixation over L2-L4 first with subsequent anterior corpectomy, cage implantation, and anterior lumbar interbody fusion was performed to achieve total tumor removal and stabilization. Pathology revealed a low grade CS with free resection margin. The patient's symptoms improved after the operation, and no recurrence had occurred as of the two-year follow-up. Furthermore, we also analyzed the post-operative outcomes of 24 cases from previous publications and our current case.

## Conclusions

Two-stage en bloc surgery concomitantly achieves total resection of the tumor, the preservation of neurological function, and a gain in stabilization. The analysis of cases showed that two-stage surgery, in comparison with one-stage surgery, seems to be beneficial in treating L-spine multiple segment disease, providing a lower rate of recurrence.

## Introduction

A chondrosarcoma (CS) is a malignant tumor comprised of transformed cells producing a cartilaginous matrix,<sup>1</sup> and such tumors account for about 10% of all bone tumors. However, primary spinal CS is relatively rare, with a reported prevalence rate of less than 10%.<sup>2</sup> Up to 60% of spinal CSs occur over the thoracic spine, with the remaining incidences variously occurring over the cervical spine, lumbar spine, and sacrum.<sup>3,4</sup> The most common symptom is back pain, along with neurological signs related to spinal cord or nerve root compression.<sup>3</sup> A palpable mass has also been mentioned to occur in cases of outward extended spinal CS.<sup>2</sup> Computed tomography (CT) usually reveals a lytic, destructive lesion of varying density, and is helpful in defining the tumor location and characterizing the tumor growth. To estimate the extent of soft tissue invasion, magnetic resonance imaging (MRI) is used. The T1-weighted images often demonstrate a hypointense lesion, whereas T2-weighted images are hyperintense.<sup>3</sup> The World Health Organization grading system for CS ranges from low-grade (grade I) to high-grade (grade IV) tumors, with the grading determined according to histologic features, such as tumor cellularity, nuclear atypia, stromal content (i.e., chondroid or myxoid), and mitoses.<sup>5</sup> CSs can be divided into conventional and variant subtypes, including clear cell CSs, mesenchymal CSs, and dedifferentiated CSs. The last two subtypes indicate high malignancy and a poor prognosis.<sup>2</sup> Taking a biopsy before definite surgery has been suggested in some articles,<sup>2,6</sup> and CT-guided biopsies performed with a large-bore trocar instead of a fine needle have also been suggested. The biopsy path should be contained in the excision margin in case of contamination.<sup>2</sup> Surgical resection is the definitive treatment for a spinal CS, with en bloc resection without entering the tumor capsule contributing to better overall survival and lower recurrence rates.<sup>4,6-8</sup> Other risk factors related to survival rates are age, histologic subtype, tumor grade, tumor size, and the extent of the disease.<sup>1,9</sup> Since spinal CS is resistant to chemotherapy and conventional radiation therapy, adjuvant radiation therapy is used for any residual tumor after incomplete resection, and has been found to improve the prognosis of metastatic patients.<sup>1,4</sup> The overall survival rate varies across different histological gradings. The 10-year survival rates are 90% and 30 ~ 40% for Grade I CS and high-grade CS, respectively.<sup>5</sup>

Several surgical approaches for achieving en bloc resection have been mentioned in previous studies. Both one-stage surgery (using only a posterior or anterior approach) and two-stage surgery (using a combination of posterior and anterior approaches) have been

mentioned. Since the choice of surgical approach for spinal CS is controversial, we present a case of primary lumbar CS along with a literature review and try to analyze the pros and cons of different approaches.

## Case Report

A 30-year-old male presented with low back pain radiating to the left anterior thigh for months. L-spine MRI revealed an L3 vertebral body osteolytic lesion with epidural expansion and compression of the spinal cord and nerve root (Fig. 1). There was no elevated tumor marker, and positron emission tomography did not reveal any abnormal uptake except in the L-spine (data not shown). Due to the remarkable neurological deficit in progression, the tumor was highly suspected of malignancy. Thus, surgical intervention was arranged.

A two-stage surgery was performed. Posterior laminectomy of L3 with transpedicular screw fixation over L2 and L4 was performed first (Fig. 2A and B). The intra-operative frozen section suggested a cartilaginous tumor (data not shown). Following the first stage of the surgery, anterior corpectomy at L3 was performed, and subsequent anterior lumbar interbody fusion (ALIF) was performed with implantation of a distractible titanium cage as well as anterior plate fixation using 76-mm VECTRA-T® (Fig. 2C and D), achieving both total resection of the tumor and reconstruction.

The pathology revealed lobules of the cartilaginous tumor with blue-grey cartilage matrix, focal increased cellularity, and minimal to focal mild nuclear atypia, suggesting the diagnosis of a grade I CS with free tumor margins (Fig. 3).

The patient's symptoms improved after the operation. The patient also reported an improved quality of life after the surgical intervention. No evidence of skeletal metastasis was found in the rest of the body in Tc-99m MDP whole body bone scan. No recurrence was noted in MRI or CT as of the two-year follow-up (Figs. 4 and 5).

## Literature Review And Analysis

A literature search was performed on August 1st, 2020, of the PubMed, Medline, and Cochrane Library databases, with the search covering studies published from 1990 to 2019. The eligibility criteria specified studies and case reports regarding patients with primary intra-osseous CSs involving the L-spine who received surgical interventions. Articles including descriptions of the surgery methods, follow-up time, recurrence, and outcomes were further selected and investigated manually. We searched the databases using three keywords, namely, "primary", "chondrosarcoma", and "spine", filtered with the race of Human, which resulted in 160 articles being identified. We next conducted a screening of the abstracts of those articles, excluding those articles in which the lesion did not involve the L-spine, those not about surgical interventions, or those in which the tumors were extra-osseous. After reviewing the full-text articles, we further excluded some articles or cases due to the disease locations, distinct tumor types, or incomplete descriptions of the surgical methods used. As a result, 8 articles discussing 23 cases were selected.

The characteristics of the selected cases, including the present case, are listed in Table 1. The average age of the patients was 39.42 ± 19.8 year-old (Mean ± SD), with 13 being male and 11 being female. The most common CS locations were L4 and L5, both of which accounted for 20.8% of the total cases.

Table 1  
List of primary intra-osseous lumbar CS cases receiving surgical intervention

No	Article	Location	Age	Sex	Staging (Enneking)	OP method	Reconstruction	Adjuvant therapy
1	Gösling et al., 2013 <sup>10</sup>	T11-L1	43	M		multilevel en bloc spondylectomy with resection and replacement of the aorta in two-stage surgery	yes, mesh	no
2	Boriani et al., 2000 <sup>11</sup>	L4	44	F	IB	en bloc	no	no
3		L4	20	M	IB	en bloc	no	no
4		L3	34	M	IB	en bloc	no	no
5		L2	31	F	IB	en bloc (corpectomy with anterior spinal fusion)	yes, ISOLA® and carbon fiber prosthesis	no
6		L5	64	M	IIB	en bloc	yes, ISOLA® 1 auto-graft	no
7		L3	68	F	IB	en bloc	yes, ISOLA® 1 carbon fiber prosthesis	no
8		L5	32	F	IB	en bloc	yes, ISOLA® 1 auto-graft	no
9		L2-L3	41	M	IIB	en bloc	yes, ISOLA® 1 CFP	PBth
10		L4	23	F	IB	piece meal excision	no	RT
11		L2-L3	33	M	IB	piece meal excision	no	RT
12		L2	56	M	IIB	piece meal excision	no	no
13		L5	21	M	IB	piece meal excision	no	no
14		L2-L4	42	M	IB	piece meal excision	no	no
15		L3	78	F	IB	piece meal excision	yes, VSP	RT
16		Hsu et al., 2011 <sup>12</sup>	T12-L2	52	M		two-stage en bloc (posterior laminectomy with TPS fixation, then anterior spondylectomy)	yes, distractible titanium cage
17	Marmor et al., 2001 <sup>13</sup>	L4	64	F		two-stage en bloc (posterior laminectomy TPS then anterior lateral spondylectomy)	yes, methylmethacrylate cage	no
18	Matsuda et al., 2006 <sup>14</sup>	L1-L2	44	M		two-stage en bloc (laminectomy and T11-T12-L3-L4 fixation, then anterior lateral spondylectomy of L1-L2 and partial T12 with titanium mesh cage)	yes, mesh	no
19	Schoenfeld, et al., 2012 <sup>15</sup>	L	64	F	IB	en bloc (by posterior and anterior approach with posterior instrumentation)		RT (and intraoperative RT)

No	Article	Location	Age	Sex (years)	Staging (Enneking)	OP method	Reconstruction	Adjuvant therapy
20		L	40	F	IIB	intralesional excision		RT
21	Tasdemiroglu et al., 1996 <sup>16</sup>	L1-L2	1	F		laminectomy and subtotal resection	no	RT and CT
22		L5	12	F		laminectomy and gross total resection with TPS fixation	no	RT and CT
23	Zibis AH et al., 2010 <sup>17</sup>	L5	9	M		two-stage en bloc (posterior laminectomy with TPS fixation, then paramedian retroperitoneal anterior approach corpectomy)	yes, titanium cage and bone graft	Neoadjuvant and adjuvant CT, and RT
24	Current case	L3	30	M	IB	two-stage en bloc (posterior laminectomy with TPS fixation, then anterior spondylectomy)	yes, titanium cage and plate	no

Table 1  
List of primary intra-osseous lumbar CS cases receiving surgical intervention (continued)

No	Article	Intralesional	Patho grade	Histologic type	Follow-up (mo)	Recurrence (mo)	Complication	Survival
1	Gösling et al., 2013	yes	low	Conventional	48	no	transient cord injury	NED
2	Boriani et al., 2000	no		Conventional	207	yes (48)		NED
3		no		Conventional	169	no		NED
4		no		Conventional	163	no		NED
5		yes		Conventional	60	yes (30)		DOD
6		no		Conventional	45	no		DUD
7		no		Conventional	45	no		NED
8		no		Conventional	30	no		NED
9		yes		Conventional	30	yes (12)		NED
10		yes		Conventional	80	yes (22)		DOD
11		yes		Conventional	119	yes (37)		DOD
12		yes		Conventional	3	progression		DOD
13		yes		Conventional	2	progression		DOD
14		yes		Conventional	36	yes (15)		DOD
15		yes		Conventional	10	progression		DOD
16		Hsu et al., 2011	yes		Clear cell	24	no	
17	Marmor, et al., 2001	no			6 (days)	no		NED
18	Matsuda et al., 2006	no		Mesenchymal	60	no		NED
19	Schoenfeld, et al., 2012	no			72	no		NED
20		yes			24	yes (24)		DOD
21	Tasdemiroglu et al., 1996	yes		Poorly differentiated mesenchymal	20			under CT
22		no		Mesenchymal	20	no		NED
23	Zibis AH et al., 2010	yes		Mesenchymal	108	no		NED
24	Current case	no	low	Conventional	12	no		NED

PBth, proton beam therapy; RT, radiotherapy; CT, chemotherapy; NED, no evidence of disease; DOD, death of disease; DUD, death unrelated with disease.

Further analysis was performed, and the results are shown in Table 2. Among the 16 patients who underwent en bloc surgery, 7 received a two-stage surgery, while 9 received a one-stage surgery. The piece meal/subtotal group had a significantly lower survival rate ( $p < 0.001$ , using Chi-square test). The recurrence rate of that group was also higher (87.5%) than that of the en bloc group (18.8%). Two patients in the two-stage en bloc group received adjuvant chemotherapy due to the use of an intralesional approach or a contaminated margin, whereas 2 patients in the one-stage en bloc group received adjuvant chemotherapy according to the chemotherapy guidelines for pediatric sarcoma. The results showed a better prognosis in general following en bloc surgery, a finding which was compatible with those of previous studies.<sup>4,6-8</sup>

Table 2  
Analysis of primary intra-osseous lumbar CS cases receiving surgical intervention

Intervention Type	Case number	Age (Mean ± SD)	Mean f/u time (mo)	Single location	Multiple location	Adjuvant Therapy	IL	Patho	Survival	Recurrence	
En bloc	all	16	40.75 ± 18.5	68	12	4	4	5		3	
	two-stage	7	43.71 ± 19.5	45	4	3	2	4	2m, 1cc, 2c	all NED	0
	one-stage	9	38.44 ± 18.4	85	8	1	2	2	1cc, 8c	1 DOD; 1DUD; 7NED	3
<b>Subtotal/Piece</b>	8	36.75 ± 23.4	37	5	3	5	8	1m, 2c	7 DOD, one under CT	7 progressed	
IL, intralesional; m, mesenchymal; cc, clear cell; c, conventional; NED, no evidence of disease; DOD, death of disease; DUD, death unrelated with disease; CT, chemotherapy											

Nearly half of the patients in the en bloc surgery group received a two-stage surgery (43.7%), including the patient in our case. Patients with multiple segment disease were inclined to receive two-stage approaches (75%, 3 of 4 patients). In contrast, one-stage approaches were preferred for single segment lesions (66.7%, 8 of 12 patients). All the subtotal/piece meal (100%) approaches were intra-lesional; however, only 31.2% of the en bloc cases were treated using an intra-lesional route (2 one-stage and 4 two-stage cases).

The most common pathological type of osseous CS among the collected cases was the conventional type, which accounted for 50% of all the cases. Notably, 8 of 9 (88.9%) one-stage en bloc cases were the conventional type. However, some of the studies and case reports did not have any description of the histologic type of the CSs in question. The mean follow-up time for the two-stage group was 45 months, while that for the one-stage group was 85 months. There was no recurrence noted in the two-stage group, while the recurrence rate in the one-stage group was 33% (3 of 9 patients, with one death from the disease). However, there was no significant difference in the recurrence rate between these two groups ( $p = 0.09$ , Chi-square test). All of the en bloc cases (2/2) with the mesenchymal type of CS underwent a two-stage surgery but no subsequent recurrence or death occurred.

## Discussion And Conclusions

Previous review studies have indicated that en bloc surgery results in better prognosis in general compared with piecemeal resection or other surgical methods, with the avoidance of intralesional contamination also being mentioned.<sup>1,4,7</sup> However, increased blood loss, prolonged operation duration, and the loss of stability owing to extensive surgical resection also cannot be ignored.<sup>4,7</sup> In the present case, we performed a two-stage surgery to achieve total resection of the tumor, to preserve neurological function, and to enhance stabilization. No sign of recurrence was noted in the described case as of the two-year follow-up.

Our analysis of studies from the last 30 years showed that in contrast to one-stage surgery, two-stage surgery seems to be beneficial in treating multiple segment disease, providing a lower rate of recurrence. Hence, two-stage surgery may provide an adequate method for treating low-grade tumors, tumor subtypes with better prognosis, or tumors with extensive invasion. However, there is still room for further discussion concerning the most appropriate choice of surgical method. The histological type and grade of CS were not well recorded in all of the literature that we reviewed. Relatedly, evidence based on histological classifications might be helpful in building up a decision tree to help CS patients achieve better prognoses and fewer complications.

Primary spinal CS is rare. En bloc resection and avoiding the use of intralesional curettage can lower the associated recurrence rate and mortality, and enhance overall survival. However, multiple factors have been identified as adversely affecting survival rates, including older age, higher tumor grade, inadequate surgical margins, and local recurrence.

The case presented herein demonstrates a promising method of two-stage surgery for an extensively invasive tumor, with the method used providing good post-operative stability and good recovery without recurrence.

## List Of Abbreviations

CS: Chondrosarcoma

CT: Computed Tomography

MRI: Magnetic Resonance Imaging

ALIF: Anterior Lumbar Interbody Fusion

## Declarations

### *Ethics approval and consent to participate*

Not applicable.

### *Consent for publication*

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editors-in-Chief of this journal

### *Availability of data and materials*

The datasets used and/or analyzed in the current study are available from the corresponding author upon reasonable request.

### *Competing interests*

The authors declare that they have no competing interests.

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### *Authors' contributions*

CDC designed the study. HRJ acquired the patient's data. YPC, CDC analyzed and interpreted the data and drafted the manuscript. CHT and CDC revised the manuscript critically for important intellectual content. All authors read and approved the final manuscript.

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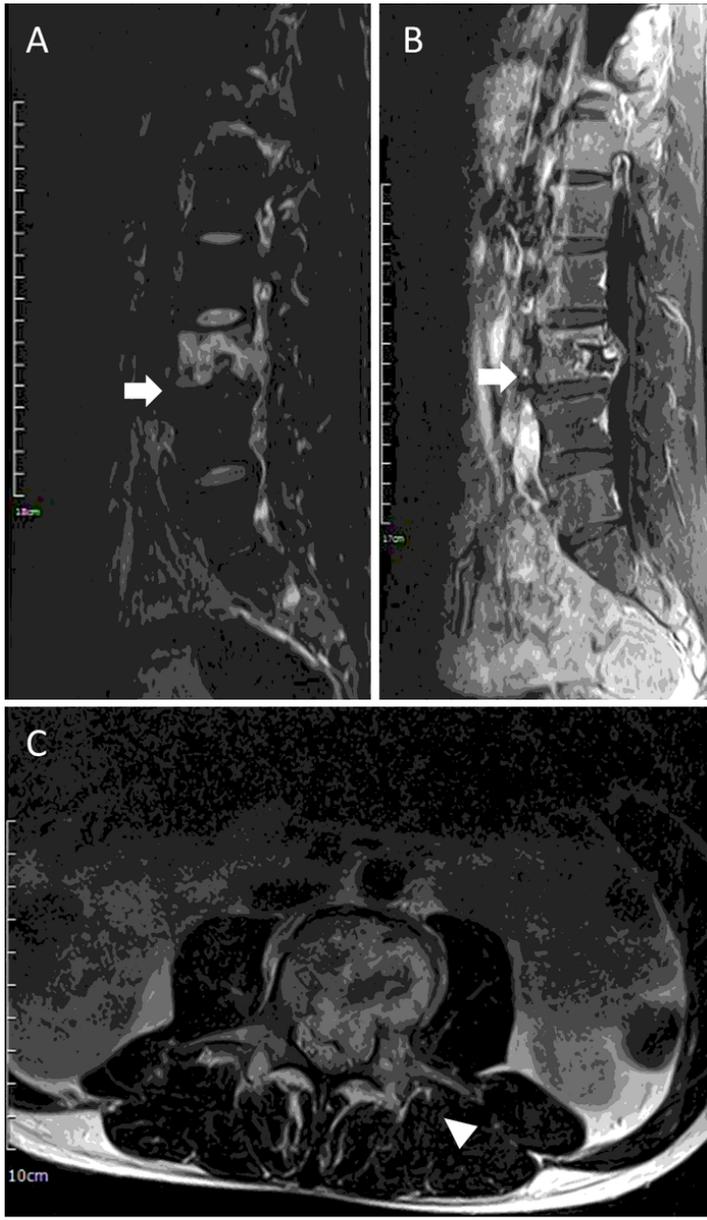
Not applicable.

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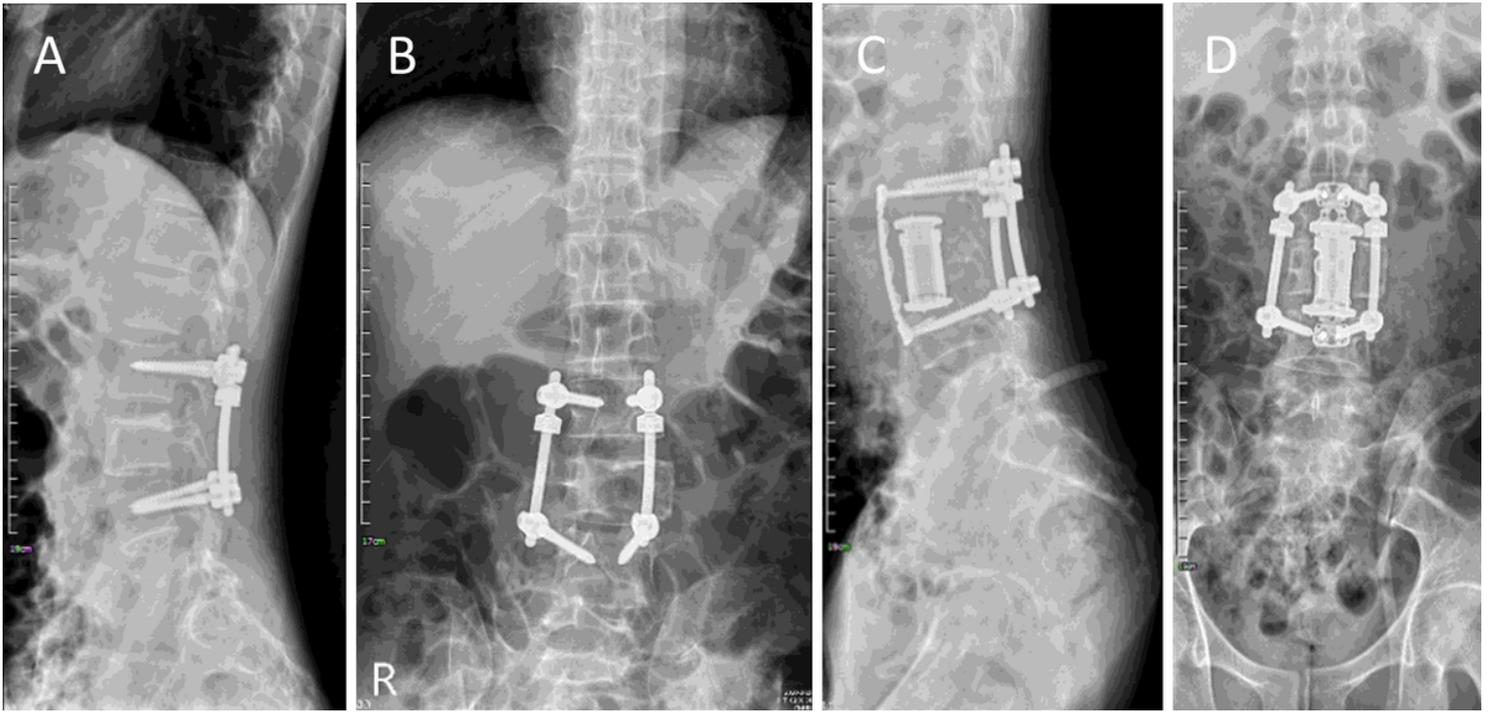
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## Figures



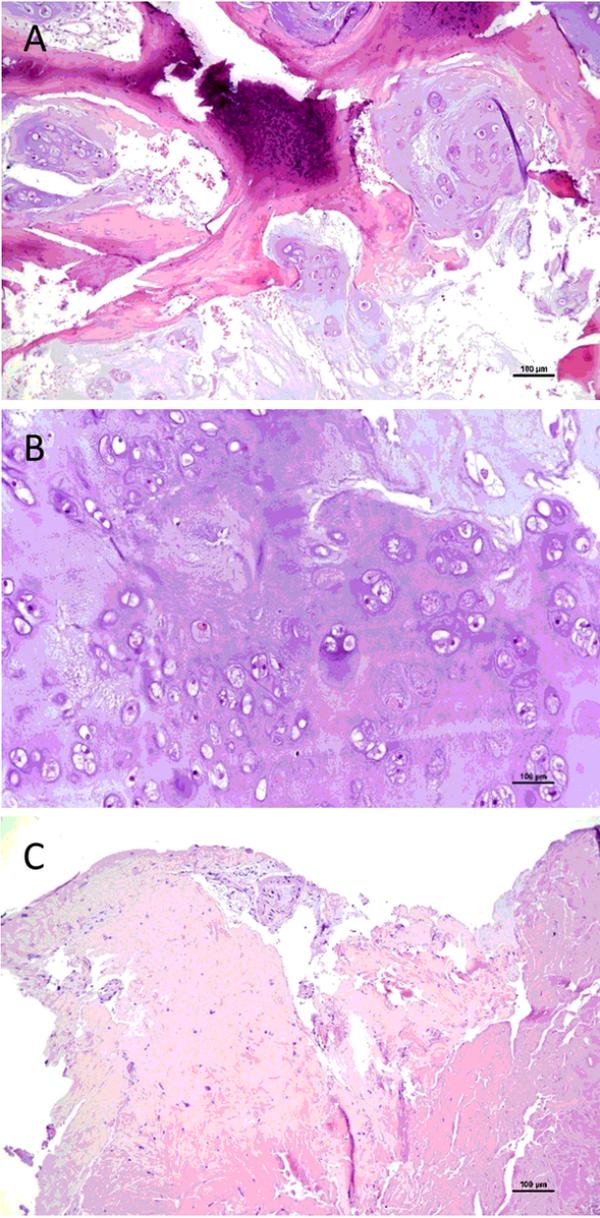
**Figure 1**

MRI scans demonstrated the lesion of the L3 vertebral body. (A) Sagittal T2-weighted image showing a lobulated lesion with heterogeneous signal intensity (arrow). (B) Sagittal T1-weighted image with contrast showing a lesion with heterogeneously high signal intensity (arrow). (C) Axial T1-weighted image with contrast showing epidural expansion of tumor associated with cord and root compression (arrowhead).



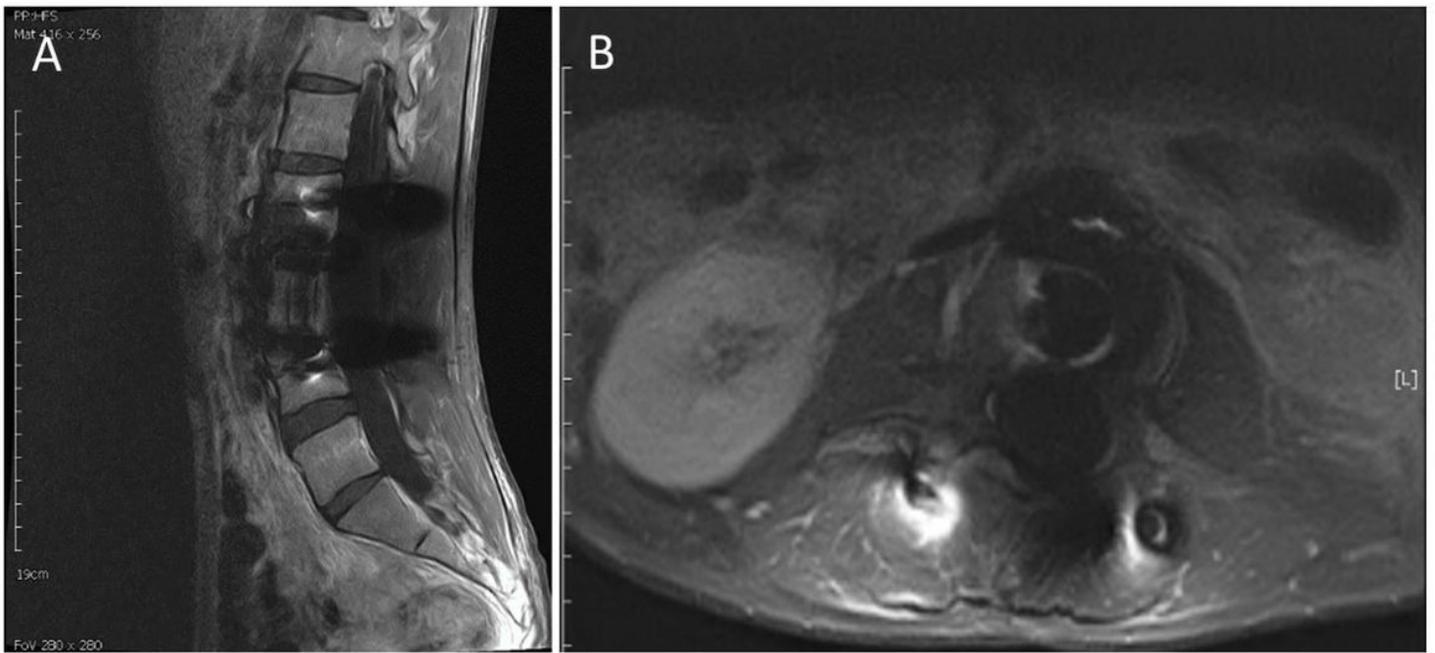
**Figure 2**

Post-operative plain films of the two-stage surgery. The lateral view (A) and anterior-posterior (AP) view (B) of L-spine after first-stage surgery showing L2 and L4 transpedicular screw fixation. The lateral view (C) and AP view (D) showing reconstruction and ALIF after L3 corpectomy.



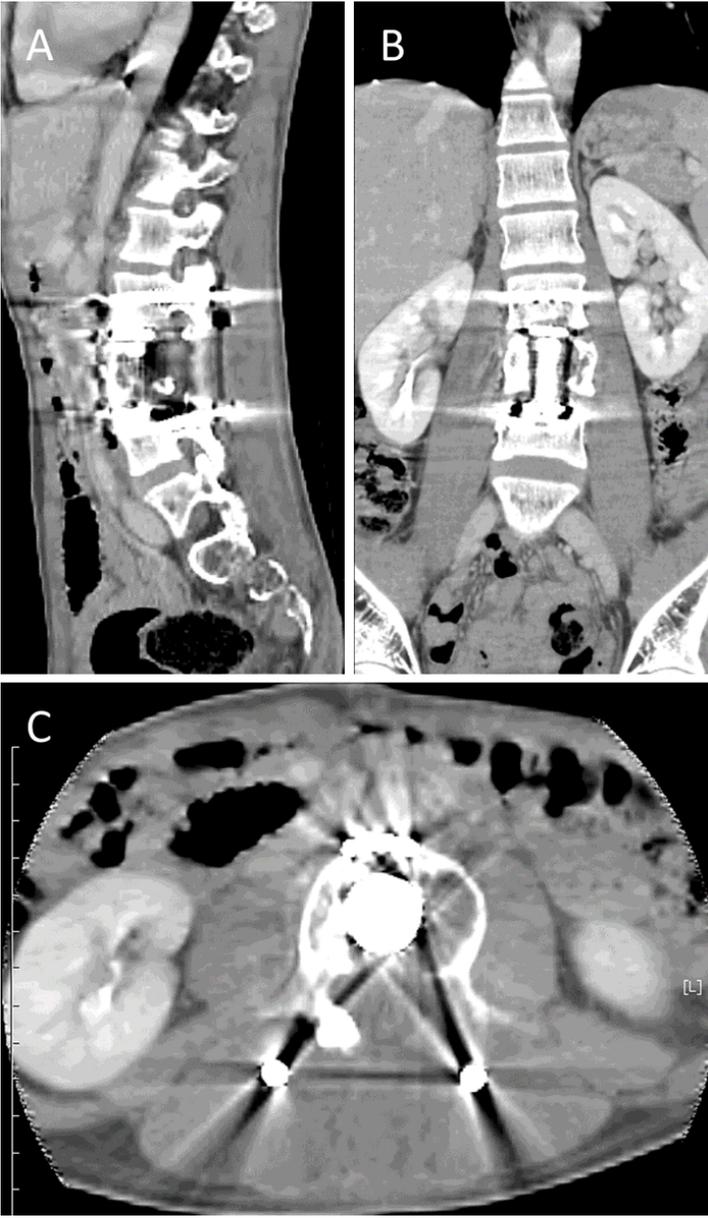
**Figure 3**

The post-operative pathological results of hematoxylin and eosin staining. 200x images showing tumor permeating and entrapping the pre-existing lamellar bone trabeculae (A) and mildly increased cellularity of the normal cartilage component (B). (C) Specimen of the margin revealing it was free of tumor. The scale bar indicates 100 µm.



**Figure 4**

One-year follow-up MRI with contrast. There was no remarkable enhancement or evidence of recurrence in the sagittal view (A) or axial view (B).



**Figure 5**

One-year follow-up CT image. There was no remarkable evidence of recurrence, with good stability of reconstruction in the sagittal view (A), coronal view (B), and axial view (C).