

Spinal dumbbell tumors: Long-term outcome and risk factors

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Abstract

Objective There is limited literature on long-term outcomes after resection of intraspinal dumbbell tumors. To identify the progression-free survival (PFS) and outcomes associated with these tumors, we retrospectively reviewed data from 74 patients.

Methods From 2007 to 2016, data from 74 patients who underwent surgical treatment for dumbbell tumors were reviewed. Patient outcomes were determined with the Japanese Orthopaedic Association (JOA) score. The median follow-up time was 7.3 years

Results Gross total resection (GTR) was performed in 39 patients (52.70%) and subtotal resection (STR) in 35 patients (47.30%). The progression-free survival (PFS) at 11 years was 82.43%. A good outcome was observed in 85.13% of patients based on the JOA. Moreover, the univariate analysis showed that surgical recession was related to tumor recurrence, and that tumor location and multiplicity were associated with tumor prognosis. However, the multivariate regression analyses showed that no factors were associated with poor prognosis and recurrence.

Conclusion The general standard treatment for spinal dumbbell tumors is complete resection, but the surgery needs to protect nerve function as much as possible. Thus, our principle is to maximize tumor removal with functional preservation.

Background

Spinal cord tumors are relatively common; the overall incidence rate is 4–16% of all central nervous system (CNS) tumors, with 13–18% being dumbbell tumors^{1–3}.

Spinal dumbbell tumors refer to tumors that connect two or more separate regions, such as intradural, epidural, and paravertebral spaces, and not just the dumbbell shape¹. The clinical manifestations of these tumors are variable depending on the location of the tumor and the involvement of various neighboring structures, but the main presenting symptom is radicular pain due to the enlargement of the tumor and thus the compression of neural structures. Due to the complexity of this anatomical structure, the importance of adjacent structures and the severity of postoperative complications, surgical treatment for the tumors is considered to be difficult. Moreover, some tumors require extensive surgical excision, such as with the use of vertebrectomy, for complete resection. There are several problems that can occur from the resection of large dumbbell tumors, including damage to the nerve root and to the vertebral artery, as well as the destruction of spinal stability, and the high recurrence rate and the risk of postoperative spinal deformities represent the major unsolved problems⁴.

Therefore, appropriate selection of the surgical approach is the key to the successful management of spinal dumbbell tumors. There have been many kinds of classifications used for dumbbell tumors in recent years, with Eden's classification and Toyama typing being the most commonly used⁵.

Due to a dearth of published reports, the effect of specific features and related recurrence factors on the outcome of patients with spinal dumbbell tumors is not extremely clear. The aim of this study was to evaluate the long-term clinical efficacy of 74 patients surgically treated for dumbbell tumors. The outcome data were then analyzed for correlations with preoperative variables, surgical morbidity, progression-free survival (PFS), resection degree, and histology, and there was a long follow-up time of 11 years.

Methods

Patients characteristics

This retrospective study of spinal dumbbell tumors included patients who received microsurgery treatment between 2007 and 2016 at our hospital. This study was approved by the Ethics Committee of our hospital and informed consent was obtained from each patient before surgery. Of 512 spinal cord tumor patients, 74 patients included in our study.

We collected the following data from each patient: the age at surgery, sex, presenting symptoms, location of the tumor (surgical level), tumor size (maximal diameter on MR images), surgical resection (gross total resection GTR or subtotal resection STR), tumor multiplicity (number of tumors), histological diagnosis, recurrence and Toyama typing. These data were assessed by the same physician. We evaluated patients' clinical symptoms and neurological function used the Japanese Orthopaedic Association (JOA)⁶. The JOA score was divided into four main categories: motor and sensory functions of the four extremities, activities of daily living (ADLs) and bladder sphincter function, with a total score of 29. All patients underwent magnetic resonance imaging (MRI) scans to determine the tumor location and size as well as the relationship with the surrounding tissue.

Surgical techniques

All patients were administered general anesthesia. Combined with preoperative X-ray, MRI and intraoperative C-arm localization, the range of the posterior median incision was determined. Posterior surgery can be used for tumors confined to the spinal canal and intervertebral foramen. The patients were placed in the prone position and the lamina and spinous process of the lesion were removed. The extradural tumors were excised first; then, the subdural part that compressed the spinal cord was excised, after which the part involved the intervertebral foramen was gradually separated to the lateral side and was excised in blocks. If exposure was limited during the operation, the osteotomy time could be extended until total resection of the tumors was complete. Most tumors often adhered to the nerve root at the injured nerve cuff; then, the tumors were dissected from the nerve root to conserve the nerve root at the lesion site as much as possible. After resection of the tumor, the field was washed with normal saline, and the dura mater was tightly sutured. Thirteen patients required spinal fixation and pedicle screws were placed in the adjacent vertebral body. Contoured rods were installed to prevent deformities. For patients with no internal fixation, a titanium plate or titanium nail was used to fix the laminae. Finally, the outer layers were sutured step by step to achieve anatomical reduction. Additionally, 9 patients underwent

second-stage thoracoscopic surgery after 3–6 months. Patients were placed in the lateral position. After ipsilateral lung collapse and single-lung ventilation, anterior resection of the paraspinal tumors was performed through a small surgical approach combined with thoracoscopy by thoracic surgeons.

Statistical analysis

All statistical data were analyzed using statistical software (SPSS Version 22.0). Categorical variables were analyzed using the Pearson χ^2 test, and the Wilcoxon rank sum test was used for continuous variables. Furthermore, we also used univariate analyses when testing for associations between outcome and recurrence. The variables included gender, age, pathology, tumor level, surgical resection, tumor multiplicity and blood loss. A P value less than 0.05 was considered statistically significant. Moreover, PFS analyses were done using Kaplan-Meier curves, measuring survival from the time of surgery to tumor recurrence.

Results

Patient demographics

Of 512 patients surgically treated for spinal cord tumors during the study period, 74 patients were included in this study. Table 1 summarizes the patient characteristics. The mean age at surgery was 45.7 years (range 10–78 years), with a slight male predominance (M/F = 1.05). The major presenting symptoms were pain, numbness, muscle weakness and sphincter disturbances, of which the most common symptom was pain (62.16%). Furthermore, the most pathological type of tumor was Neurilemmoma (63.51%) and most tumors were located in the cervical region (39.19%). Moreover, tumor sites and distributions are also summarized in Table 1. The median follow-up time was 7.3 years (range 3 – 11).

Table 1
Patients characteristics

Patient's characteristics	Number	Percent of 51
Number of patients	74	100%
Sex		
male	38	51.35%
female	36	48.65%
Presenting symptoms		
pain	46	62.16%
numbness	45	60.81%
muscle weakness	37	50.00%
sphincter disturbances	9	12.16%
Location of the tumor		
Cervical	29	39.19%
Thoracic	28	37.84%
Lumbosacral	10	13.51%
Cervicothorax or Thoracolumbar	7	9.46%
Pathology		
Neurilemmoma	47	63.51%
Melanotic schwannoma	2	2.70%
Meningioma	5	6.76%
Neurofibroma	9	12.17%
Ganglioneuroma	3	4.05%
Hemangiopericytoma	1	1.35%
Hemangioblastoma	1	1.35%
Hemangioma	4	5.41%
Multiple myeloma	1	1.35%
Chondrosarcoma	1	1.35%
Tumor multiplicity		

Patient's characteristics	Number	Percent of 51
Single	68	91.89%
Two or more	6	8.11%
Tumor size		
More than 4 cm	13	17.57%
Less than 4 cm	61	82.43%
Surgical resection		
GTR	39	52.70%
STR	35	47.30%
Internal fixation		
Yes	13	17.57%
No	61	82.43%
Postop improvement rate (JOA)		
75–100%	25	33.78%
50–74%	38	51.35%
25–49%	7	9.46%
0–24%	4	5.41%
Tumor recurrence		
yes	13	17.57%
no	61	82.43%

Tumors and surgery

The dumbbell tumors were divided into 9 categories according to Toyama classification. The detailed classification information is shown in Table 2, where the most prevalent type was IIIA (58.10%). Of the 74 patients, GTR was performed in 39 patients (52.70%), and STR was performed in 35 patients (47.30%). Thirteen patients underwent spinal pedicle screw fixation or bone graft fusion and internal fixation. In terms of the reason for STR, as summarized in Table 2, the most reason was that the tumor extended too far into the intervertebral foramen, thus leading to incomplete exposure (28.57%) followed by postoperative complications (9/74, 12.16%). Six patients had chemical meningitis (6/74, 8.10%) and recovered with antibiotics. Three patients developed impairments in wound healing (3/74, 4.05%). No patients displayed postoperative vertebral artery injury, spinal instability, motor disorders or sensory disturbances.

Table 2
Toyama typing and the reason of STR

	Number	Percent
Toyama typing		
I	5	6.76%
II	11	14.86%
A	8	
B	3	
C	0	
III	54	72.97%
A	43	
B	11	
C	0	
IV	0	0
V	3	4.05%
VI	1	1.36%
The reason of STR		
Extending too far into the intervertebral foramen and incomplete exposure	10	28.57%
Enveloping vertebral artery	6	17.14%
Vertebral venous plexus caused excessive bleeding	6	17.14%
Nerve roots cannot be separated from tumors	6	17.14%
Hemorrhage of blood supply arteries of tumors	1	2.86%
Closely related with thoracic or descending aorta	2	5.71%
Invasion of surrounding tissues	1	2.86%
The widespread of tumor basement or widely distributed	3	8.57%

Follow-up and analysis of prognostic factors

We used the JOA score to access neurological function. The recovery rate was calculated as follows: $(\text{postoperative JOA score} - \text{preoperative JOA score}) / (\text{full score} - \text{preoperative JOA score}) \times 100$. The rate was considered excellent (75–100%), good (50 – 74%), fair (25 – 49%), or poor (0–24%). Thus, a rate higher than 50% was identified as a good outcome. The mean preoperative and postoperative JOA scores

were 18.99 ± 3.43 and 23.02 ± 4.21 , respectively. Specifically, there was a significant difference between the preoperative and follow-up JOA scores ($P < 0.001$). All in all, 85.13% (63/74) of patients had a good outcome.

Furthermore, the progression-free survival (PFS) in our study was 82.43% (Fig. 1). Thirteen patients (17.57%) experienced tumor recurrence or progression of residual tumor. Ten patients underwent a second operation to treat recurrent tumors, and the rest did not undergo an operation because they could not tolerate general anesthesia. Tumor recurrence might have been associated with STR or malignant tumors. During our follow-up period, 7 patients died because of advanced age and other internal diseases.

To explore the variables possibly associated with prognosis and recurrence, we adopted both univariate and multivariate regression analyses (Tables 3 and 4). Univariate analysis showed that surgical recession was related to tumor recurrence and that tumor location and multiplicity were associated with tumor prognosis. However, the multivariate regression analyses showed that no factors were associated with poor prognosis and recurrence.

Table 3

Univariate analyses of variables possibly associated with recurrence and prognosis

	Recurrence			Prognosis		
	Yes	No	P value	Good	Poor	P value
Sex	0.306			0.377		
Male	5	33		31	7	
Female	8	28		32	4	
Age	0.710			0.901		
≤60	12	51		53	10	
>60	1	10		10	1	
Tumor location	0.302			0.003		
Cervical	3	26		27	2	
Thoracic	5	23		25	3	
Lumbosacral	2	8		9	1	
Cervicothorax or Thoracolumbar	3	4		2	5	
Tumor size	0.820			0.359		
More than 4 cm	2	11		10	3	
Less than 4 cm	11	50		53	8	
Surgical resection	0.018			0.067		
GTR	3	36		36	3	
STR	10	25		27	8	
Blood loss	0.929			0.525		
≥800	2	10		9	3	
<800	11	51		54	8	
Tumor multiplicity	0.618			0.001		
Single	11	57		62	6	
Two or more	2	4		1	5	
Tumor properties	0.281			0.191		
Benign	11	59		61	9	

	Recurrence		Prognosis	
malignant	2	2	2	2
The level of tumor	0.641		0.930	
More than C4	3	18	18	3
Lower than C4	10	43	45	8

Table 4

Multivariate regression analyses of variables possibly associated with prognosis and recurrence

Influence factor	Prognosis		Recurrence	
	P value	Odds ratio (Confidence interval of 95%)	P value	Odds ratio (Confidence interval of 95%)
sex	0.528	0.544(0.082–3.597)	0.936	0.945(0.241–3.709)
age	0.999	—	0.999	—
The level of tumor	0.298	0.056(0.00-12.78)	0.999	—
Tumor size	0.316	4.222(0.253–70.46)	0.599	0.623(0.107–3.636)
Tumor location	0.998	—	0.782	0.909(0.465–1.781)
Tumor properties	0.999	—	0.999	—
Surgical resection	0.703	1.464(0.206–10.377)	0.410	0.560(0.141–2.227)
Tumor multiplicity	0.999	—	0.999	—
Blood loss	0.999	—	0.808	1.263(0.193–8.244)

Discussion

Previous studies have described the incidence of spinal dumbbell tumors as 13–18%^{1–3}. In our study, the rate was 14.45%, which is consistent with these reports. Therefore, dumbbell tumors are not unusual. Moreover, many studies have reported that dumbbell tumors occur most commonly in the cervical spine and that most are neurilemmoma^{1,7–9}. Our research showed that 63.51% of tumors were neurilemmoma, but there was no significant difference in the location of tumors between the cervical and thoracic segments. Of the 74 tumors, the following 4 were malignant (5.40%): hemangiopericytoma, hemangioblastoma, multiple myeloma and chondrosarcoma. This incidence was slighter lower than the

incidence found in previous studies^{1,10}. Moreover, the clinical symptoms depended on the location of the tumor and there was no obvious specificity, with symptoms including pain and weakness of the lower limbs.

In 1941, Eden was the first to classify spinal dumbbell tumors and this classification was widely recognized. However, due to the backwardness of medical technology at that time, this classification system did not clearly help with preoperative judgment of the relationship between tumors and adjacent structures, especially in the formulation of surgical strategies. In 2004, Asazuma et al. proposed a new classification system with nine categories called Toyama typing⁵. This approach was the first classification based on three-dimensional computed topography (CT) or magnetic resonance imaging (MRI). Because this classification is more explicit and convenient than Eden's classification for our study, we adopted this classification for our research. Moreover, there have been other classifications proposed for dumbbell tumors. Jiang et al. developed a new classification system that included 7 categories (types 1–7) and 2 foraminal modifiers for determining the surgical approach⁴. In addition, Liu et al. recommended a novel classification of spinal dumbbell tumors based on the characteristics of the surgical approach⁸. Their study aimed to help the surgeon select a suitable surgical approach. Regretfully, this was a retrospective study of a single surgeon's experience and the validity has not been confirmed by large-scale samples or other scholars. To date, operative approaches to dumbbell tumors are still controversial, but the choices are mostly based on the surgeon's familiarity with regional anatomy and on personal preference.

An ideal surgical strategy for dumbbell tumors should be able to reduce the recurrence rate and simultaneously avoid spinal deformities. All kinds of surgical approaches for dumbbell tumors including total resection of spinal dumbbell tumors have been implemented, for instance, a single posterolateral or anterolateral approach, a combined posterior and anterior approach and an anterolateral approach with the use of 2 stages. We believe that most dumbbell tumors can be satisfactorily resected with the single posterior approach; McCormick has also advocated that dumbbell tumors can be effectively managed with a single posterior approach combined with laminectomy¹¹. Some scholars have reported good results with the use of an anterior approach for the total removal of cervical dumbbell tumors¹². Lot and George used a lateral approach and vertebral artery ligation for complete resection¹³. In addition, Jiang et al. treated patients with an anterior followed by a posterior approach for gross total resection, but found a high rate of complications⁴. However, most surgeons are unfamiliar with the anatomy associated with these approaches. More importantly, tumor removal with the use of these approaches is associated with a risk of injuring the vertebral artery (VA) and the accessory or hypoglossal nerves. Yong huang et al. suggested that a combined anterior and posterior approaches only be performed when there is residual tumor, but this suggestion was not widely accepted by patients and required a long surgical time¹⁴. However, the posterior approach is popular and safe and has a low potential for damaging important structures. As long as the VA can be visualized and protected, a one-stage posterior approach can be performed and even extended to the ventral side of the vertebral artery¹⁴. For these reasons, we prefer the posterior approach for our patients.

In addition, for cervical dumbbell tumors, attention should be paid to the protection of the vertebral artery. There is usually a capsular layer between the neurilemmoma and the vertebral artery. As long as the tumors do not completely encapsulate the vertebral artery, most of them can be completely separated from the vertebral artery. In addition, when approaching the vertebral artery, venous hemorrhage is often obvious. Most of these cases are hemorrhage of the venous plexus around vertebral artery. For thoracic dumbbell tumors, most scholars have suggested a combined approach followed by thoracoscopic removal of the intrathoracic component is the standard procedure^{15,16}. However, Y. Li et al. demonstrated that the posterior approach achieved better results and fewer complications compared to the combined surgical approach and they thought the posterior approach allowed for the interaction with familiar structures throughout the operation, requiring only a single incision and avoiding the need for a postoperative chest tube, which diminish potential complications such as postoperative pain, pulmonary dysfunction and infection¹⁷. In our study, 9 patients underwent second-stage thoracoscopic surgery because of large paravertebral tumor and none experienced complications after surgery.

Generally, GTR of spinal dumbbell tumors is ideal because it reduces the chance of recurrence. Some studies have reported an increased recurrence rate after STR and have strongly suggested complete resection^{1,4,18}. Moreover, Klekamp et al. showed a recurrence rate of 10.7% at 5 years and 28.2% after 10 to 15 years in patients¹⁹. However, Jiang et al. suggested the recurrence rate of patients with GTR was only 2.8% with an average follow-up of 5years⁴. One of the reasons for the high recurrence rate is the tumor capsule. As Benzel et al. reported removing nerves from the tumor origin rather than nenucleation, can avoid the recurrence of tumors²⁰. The other reason is the subtotal resection rate. Sometimes it is difficult to remove the tumor without damaging the entire nerve root, and it is even sometimes impossible to achieve radical resection. Previous studies have noted that GTR can be achieved in 86–95% of patients^{5,18,21}. However, the risk of injury is sometimes high for radical resection and 20% of patients present with radicular deficits⁴.

In our study, 52.7% of patients underwent GTR, and the reasons for STR were as follows: 10 patients' tumors extended too far into the intervertebral foramen and thus caused incomplete exposure; 6 patients' tumors were too close to or enveloped the vertebral artery to avoid unnecessary bleeding; 6 patients' tumors adhered tightly to nerve roots and were unresected; vertebral venous plexus hemorrhage resulted in unclear intraoperative visual field in 6 patients; the artery that provided blood supply to the tumor was bleeding in 1 patient; 2 patients' tumors were located close to the thoracic or descending aorta; 3 patients' tumors were widespread basement tumors or were widely distributed; and 1 malignant tumor invaded the surrounding tissues. However, only 17.57% of patients had tumor recurrence after an average 10-year follow-up, and this result was lower than the 28.2% recurrence rate reported by Klekamp et al. after 10 to 15 years and the 20% recurrence rate reported by Sung Mo Ryu et al. after 35-month follow-up^{19,22}. In addition, only 9 patients had complications, but no major complications were observed and good neurologic outcomes were achieved. In a recent study, Sung Mo Ryu et al. noted that the overall neurologic outcomes were better in the STR group than in the GTR group²². Thus, we aimed to maximize tumor removal with functional preservation.

The PFS of our study was 82.43%, Based on the JOA score (50–100%), 85.13% of patients had a good outcome, which was slighter higher than the outcomes reported by K. Ito et al. and Sung Mo Ryu et al.^{22,23}. Thus, it is significant to understand the risk factors for tumor recurrence and prognosis. The univariate analysis showed that recurrence and poor outcome were related to STR and tumor location, respectively ($P < 0.05$). However, multivariate regression analyses did not show any positive results. The difference between the two analyses may be due to the limitation of the small sample size in our study.

Conclusion

Data from seventy-four patients with spinal dumbbell tumors were reviewed. The PFS at 11 years was 82.43% and a good prognosis was considered in 85.13% of patients based on the postoperative JOA score. The gold standard treatment for dumbbell tumors is GTR, but the premise is to ensure good neurologic outcomes. It is noteworthy that this study had limitations because this was a retrospective follow-up study, and we had a sample size of only 74 dumbbell tumors, which may affect the credibility of our findings. We believe that our findings may have significant clinical implications in terms of spinal dumbbell tumors and may provide a basis for future studies. Moreover, considering the characteristics of these tumors, further studies are needed in the long term.

Declarations

Abbreviations

PFS: progression-free survival

JOA: the Japanese Orthopaedic Association score

GTR: gross total resection

STR: subtotal resection

MRI: magnetic resonance imaging

ADL: activities of daily living

CNS: central nervous system

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Availability of data and materials

None.

Author contributions

JG and XW designed the research; XW, YL, SH and ZL performed the research; XW and YL collected the data; XW and JG analyzed and wrote the paper; Besides, all authors reviewed and commented on the manuscript.

Ethics approval and consent to participate

Our research was retrospective study which is not applicable for consent.

Consent for publication

The manuscript has not been published previously, in any language, in whole or in part, and is not currently under consideration elsewhere.

Competing interests

The authors declare that they have no competing interests.

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Figures



Figure 1

Graph showing Kaplan–Meier estimates of PFS.