Original Article

Clinical features associated with recurrence of tumours of the spinal cord and cauda equina

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Study design: Retrospective review of consecutive cases of recurrent spinal cord and cauda equina tumours.

Objectives: We sought to identify factors and conditions resulting in re-operation to treat recurrences of spinal cord and cauda equina tumours.

Setting: Keio University Hospital, Tokyo, Japan.

Methods: Re-operation was performed in 39 patients with spinal cord and cauda equina tumours. Times of operation, interval between operations, affected spinal level, tumour site on cross section, configurations among dumb-bell tumours, and pathologic diagnoses were analysed. Recurrence rates were defined in terms of the number of cases with re-operation due to tumour recurrence relative to the total number of surgical cases for the same period at our institution.

Results: Recurrence rates were relatively high for intradural, extramedullary tumours and for tumours located anteriorly rather than laterally. Of patients with intradural, extramedullary plus extradural tumours who underwent initial surgery at our hospital, 75% (9/12) recurred; all tumours had dumb-bell-type configurations. The overall rate of re-operation due to tumour recurrence in 249 cases was 7.2% at our institution. By tumour types, 40% of malignant schwannomas recurred (2/5), as did 35.7% of neurofibromas (5/14), and 33.3% of ependymomas (6/18).

Conclusion: Risk factors for tumour recurrence were anterior location, an intradural, extramedullary plus extradural site, extensive dumb-bell tumours, and pathologic diagnoses of neurofibroma, ependymoma, or malignant schwannoma.

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Keywords: spinal cord tumour; recurrence; dumb-bell tumour

Introduction

The goal of the treatment for spinal cord and cauda equina tumours is to resect the lesion totally without injuring the spinal cord, cauda equina, or spinal nerve roots, or exacerbating the patients' neurologic deficit. However, we eventually need to re-operate on some patients because of tumour recurrence. Very few reports^{1–4} have described the comprehensive clinical features of patients with spinal cord and cauda equina tumours that required re-operation because of tumour recurrence. In a retrospective analysis of cases of spinal cord and cauda equina tumour, we sought to identify

factors and conditions associated with tumour recurrence necessitating a re-operation.

Methods

Between 1963 and 1994, re-operation was performed in 39 patients with recurrent spinal cord and cauda equina tumours in the Department of Orthopaedic Surgery at Keio University Hospital. Patients included 20 men and 19 women, with ages at the time of final surgery ranging from 2 to 73 years (mean, 40). The first operation was performed at our hospital for 18 patients (46.2%) and at others for 21 patients (53.8%). Using medical records including diagnostic imaging such as myelography, computed tomography

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(CT) myelography, and magnetic resonance imaging (MRI), the timing of an operation, the interval between operations, affected spinal level, tumour site on cross-section, and pathologic diagnosis were analysed. For dumb-bell tumours, the detailed anatomic configuration was also noted. Site of tumours in relation to the spinal cord were classified as anterior, anterolateral, lateral, posterolateral, and posterior (Figure 1). The configuration of dumb-bell tumours was classified according to Toyama's classification⁵ including nine types (type I to VI) and subtypes (Figure 2). Recurrent spinal cord tumour was defined as a tumour with the same histologic diagnosis as was evident at the first operation, as well as occurrence at the same spinal level.³ Recurrence rate was defined as the ratio between the number of cases with re-operation due to tumour recurrence and the total number of surgical cases at our hospital in the same period. Variables representing risk factors for tumour recurrence were determined statistically using Fisher's exact probability test.

Results

Twenty-eight patients underwent surgery twice, while 11 patients required three operations. The radicality of each surgery is shown in Table 1. Intervals between the first and second operation ranged from 6 months to 27 years (mean, 5 years and 4 months). Intervals between the second and third operation ranged from 1 year and 9 months to 19 years (mean, 5 years and 3 months).

Table 1 Tumour radicality at each surgery (No. of patients)

	Complete	Incomplete	Unknown
First surgery	0	35	4*
Second surgery	24	15**	0

*Four patients underwent first surgery at other hospitals and surgical records were not obtained.

**Four patients were dead without undergoing the third surgery

Figure 1 Tumour sites on cross-section

Between 1963 and 1994, we surgically treated 249 patients with spinal cord and cauda equina tumours. During this period, we encountered 18 patients with recurrent spinal cord and cauda equina tumours who underwent their first operation at our hospital and needed re-operation. Consequently, the rate of re-operation due to tumour recurrence (recurrence rate) was 7.2% at our hospital. The tumours were cervical in 12 patients (30.8%), cervicothoracic in four patients (10.3%), thoracic in seven patients (17.9%), at conus level in two patients (5.1%), at the cauda equina level in 12 patients (35.9%), and wide-ranging at multiple level in two patients (5.1%). The rate of recurrent cases was 16.7% (12/72) for the cervical region; 28.6% (4/14) for cervicothoracic; 10.4% (7/67) for thoracic; 11.1% (2/18) for conus level; 17.9% (12/67) for cauda equina; and 18.2% (2/11) for multiple-level involvement. No significant difference was noted between these regions.

Tumour site on cross-section as determined by diagnostic imaging and intra-operative findings was intradural and extramedullary in 17 cases (43.6%), extradural in eight (20.5%), intramedullary in five (12.8%), and intradural and extramedullary plus extradural in nine (23.1%). The rate of recurrence with respect to tumour site on cross-section considering all surgical cases was 75% (9/12) among intradural and extramedullary plus extradural tumours; 14.8% (17/115) for intradural, extramedullary tumours; 11.4% (8/70) for extradural tumours; and 11.1% (5/45) for intramedullary tumours. A total of 242 cases were considered, excluding seven cases where cross-sectional location was unknown. The recurrence rate of intradural, extramedullary tumours was higher than for the other groups ($P < 0.001$).

The extramedullary tumour site in relation to the spinal cord was identified in 21 recurrent cases (excluding cauda equina tumours): four were anterior (19.0%), four were anterolateral (19.0%), two were lateral (9.5%), four were posterolateral (19.0%), six were posterior (28.6%), and one was circumferential (4.8%). Recurrence rates considering a total of 145 surgical cases in this period where location was identified were as follows: 40% for anterior (4/10), 11.8% for anterolateral (4/34), 7.4% for lateral (2/27), 12.1% for posterolateral (4/33) and 14.6% for posterior (6/41). The rate of recurrence was higher in patients whose tumours were located anteriorly than in those with laterally situated tumours ($P < 0.05$).

Seventy-five percent of patients with intradural and extramedullary plus extradural tumours who underwent initial surgery at our hospital (9/12) represented recurrent cases, all dumb-bell type tumours. Dumb-bell tumours were observed in 15 patients (38.5%) including the 12 patients above. According to the classification of dumb-bell tumour configuration, four patients had type IIa; four, type IIIa; three, type IIb; three, type VI; and one, type IV. In these cases, tumours extended widely beyond the intervertebral

Figure 2 Classification of dumb-bell tumours according to Toyama⁵

foramen or invaded the vertebral body. Only a posterior approach was used in these recurrent cases of type IIIb cervical dumb-bell tumours.

Pathologic diagnoses of the 39 recurrent cases were neurinoma in 14 cases (35.9%), meningioma in six (15.4%), ependymoma in six (15.4%), neurofibroma in five (12.8%), malignant schwannoma in two (5.1%), lipoma in two (5.1%), and others in three (7.8%). Rate of recurrent cases among a total of 249 cases including 40% for malignant schwannoma (2/5), 35.7% for neurofibroma (5/14), 33.3% for ependymoma (6/18), 18.2% for meningioma (6/33), 13.3% for lipoma (2/15), and 12.0% for neurinoma (14/117). Recurrence rates for neurofibromas and ependymomas were significantly higher than for neurinomas ($P < 0.05$).

Microscopic surgery was introduced in 1982. Since then, microscopic surgery was done for all the patients with intramedullary tumours. Only one patient with ependymoma recurred and underwent re-operation in spite of using surgical microscope out of six re-operative patients.

Illustrative case

A 49-year-old, woman presented to another hospital with low back pain and paraparesis. Complete block was found at the T10–11 level in a descending myelogram and at the L3–4 level in an ascending myelogram. Spinal cord tumour resection was performed by a posterior approach. The pathologic diagnosis was neurinoma. Symptoms disappeared al-

most completely after the first operation. However, numbness in her lower limbs was noted 3 years postoperatively. MRI demonstrated the tumour recurrence at the L3–4 level (Figure 3a,b). The recurrence was located at the outer aspect of the vertebral foramen and in the vertebral body. The second operation was carried out at our hospital, and total removal of tumour was accomplished. The tumour was a type VI according to Toyama's classification.

Discussion

In previous reports, rates of re-operation were between 7.5 and 15.5%.^{1–4} Iwahara and Iseki¹ reported that 75% of re-operations resulted from misdiagnosis, and that most recurrence represented residual dumb-bell tumour in the epidural space. In other reports, recurrence was observed in tumours located anteriorly to the spinal cord² and at the cauda equina level.³ After CT was introduced in 1982, the re-operations for patients who underwent their first operation at our institution numbered only three. Remarkable progress in diagnostic imaging such as CT and MRI reduced the rate of recurrence attributable to macroscopically evident residual tumour. We performed tumour resection using surgical microscope in 15 patients with ependymomas since 1982. Total resection was achieved in 11 patients. Tumour recurrence was observed and re-operation was done in only one of these patients who underwent partial resection.⁶ This fact means that advances in surgical technique also contributed to the decrease in recurrence rate, especially in intramedullary tumours.

Considering recurrence rate according to the spinal level, Shimizu *et al.*³ reported a significantly higher rate for cauda equina tumours than for more rostral tumours. However, we found no significant difference between spinal cord and cauda equina level (25/116 *versus* 12/67).

Our findings identified risk factors for tumour recurrence as location anterior to the spinal cord; Intradural and extramedullary plus extradural configurations particularly dumb-bell tumours; and pathologic diagnoses of neurofibroma, ependymoma, or malignant schwannoma.

The rate of tumour recurrence was highest (40%) along the anterior aspect of the spinal cord. In cases of meningioma located at this site, some macroscopic tumour is likely to be left, and management of the dura mater is technically difficult. Gezen *et al.*⁷ emphasised technical difficulties of tumour resection, especially those involving a location ventral to the cord.

Configuration of dumb-bell tumours were mostly extradural, such as types II, III, IV and VI in Toyama's classifications.⁵ The cause of recurrence was residual tumour at the site of the foramen and anterior to it because the tumour was removed via a posterior approach at the first operation. To avoid this situation, a combined anterior and posterior approach

Figure 3 Illustrative case involving 49-year-old woman. (a) A T1-weighted, gadolinium-enhanced sagittal magnetic resonance image obtained after recurrence 3 years later shows enhancement of the tumour at the L3 level. (b) A T1-weighted gadolinium-enhanced axial image shows the enhancing tumour invading the vertebral body and foramen

should be used for dumb-bell tumours located at the site of the foramen and anterior to it, such as type IIIB. Alternatively, Lot and George⁸ recommended antero- or posterolateral approaches to achieve total tumour resection.

Among intramedullary tumours, re-operation was performed in six cases of ependymoma because of

recurrence. In our previous reports, eight of 26 patients (30.8%) who underwent partial or subtotal resection developed a clinical recurrence.⁶ Ferrante *et al.*⁹ similarly reported that eight patients among those who underwent subtotal removal, developed a recurrence, while a patient who underwent total tumour removal had a clinical recurrence. Cooper¹⁰ reported that six of 13 patients who had 99% or subtotal removal had a clinical recurrence, in contrast to only one of 11 patients with total resection. Malignant tumour types such as malignant schwannoma also represent risk factors, either because of invasiveness precluding complete resection or because of intrinsically aggressive histologic features.

The recurrence rate for completely removed meningioma has been reported to vary between 1.3 and 11%.^{11–14} The re-operation rate due to recurrence for our meningiomas (18.2% or 6/33) was relatively high because complete removal was not undertaken in some cases. The rate of clinical recurrence can be expected to be higher in partially removed meningiomas than in totally removed tumours; the difficulty of excision of the dura at the point of origin in meningioma often is cited.¹² Roux *et al.*¹⁵ described two patients who underwent subtotal tumour removal and presented with recurrences 2 years after initial surgery, despite electrocoagulation of the dural attachment. Simpson¹⁶ recommended radical resection, including surrounding dural tissue to minimize likelihood of recurrence. Klekamp and Samii¹⁴ also emphasised that except for encapsulated tumours, extended dura resections probably are required to lower the chance of recurrence. However, in another series, no recurrence was noted in 15 tumours, mostly located anteriorly, for which the dural origin was simply electrocoagulated.¹² We would often defer treatment of the dura mater in elderly patients when the meningioma is not pathologically malignant and is considered to be totally removed macroscopically, because more than 10 years would be required for possible appearance of symptoms.¹⁷ However, atypicality or malignancy of a meningioma can be difficult to assess intraoperatively. Aggressive treatment of the dura mater overlying the anterior aspect of the spinal cord should be considered in young patients or in cases where malignancy is suspected.

Analysis of configuration and location of tumours by imaging in three dimensions, as well as distinguishing between neurinoma and meningioma based on gadolinium-enhanced MRI are highly important for

surgical planning. Improved planning and optimal surgical technique are indispensable for preventing tumour recurrences requiring re-operation.

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