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### Intradural Extramedullary Spinal Cord Tumors and Outcome with Surgical Approach: A Single Centre Retrospective Study

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#### ABSTRACT

**Background:** Intradural extramedullary spinal tumor (IDEM) is rare compared to all neoplasms of the central nervous system; however, it is the most common type of primary intraspinal neoplasms. Most tumors are meningioma, schwannoma, and neurofibroma. IDEM tumor has a predilection in women with ages ranging from 40-60 years. This study was conducted to describe the outcome of patients with IDEM tumors after surgery in Dr. Moewardi General Hospital from 2019-2022. **Methods:** This retrospective descriptive study was conducted in 2019-2022. Forty-four patients diagnosed with IDEM tumors and who underwent surgical approach were included. The studied variables include age, pathological diagnosis, intervention, and postoperative follow-up. **Results:** Most subjects were within the age range of 40-60 years old (52.3%), women (63.6%), and had schwannoma (45.5%), followed by meningioma (25%). Based on gender, both men and women mostly had schwannoma (56.25% and 39.3%, respectively). All subjects underwent tumor excision laminectomy. In the follow-up, most subjects (86.4%) had improved clinical symptoms after surgery. **Conclusion:** Intradural extramedullary spinal tumors mostly occur in women 40-60 years old, and schwannoma and meningioma are the most frequent types. Laminectomy intervention can provide clinical symptom improvement in most patients.

#### 1. Introduction

Intradural extramedullary spinal tumor accounts for two-thirds of all primary intraspinal neoplasms. However, it is considered a rare tumor. Of all central nervous system tumors, approximately only 15% of tumors are intraspinal tumors. Most tumors are benign; about 60% occur in extradural, 30% intradural extramedullary (IDEM), and only 10% as the true intramedullary spinal cord tumor (IMSCT). The reported incidence of IDEM tumors is 3-10 per 100,000 population, and most arise from the nerve sheath (about 30%) and meninges (about 25%). In particular, the incidence of schwannoma is 0.3-0.4

cases per 100,000 population, and meningioma is 0.32 cases per 100,000 population annually.<sup>1-3</sup>

Based on the demographics, most IDEM tumors occur in women compared to men, and this tumor tends to occur in the fourth to fifth decade of life.<sup>4</sup> The most common primary extramedullary tumor is meningioma. Meningioma arises from the meninges arachnoidal cap. This tumor accounts for 25% of all spinal neoplasms and mostly occurs in the thoracic vertebrae and the posterolateral position. Based on the histology, meningioma can be classified as psammomatous, fibroblastic, and meningothelial. Most meningioma type is characterized by their slow

growth and benign behaviour (WHO Grade I), while tumors with more rapid growth and malignant behaviour are considered WHO Grade II and III.<sup>5</sup> The second most common extramedullary tumor is nerve sheath tumors, spinal schwannoma, or neurofibroma. Schwannoma is a slow-growing lesion and arises from the dorsal sensory roots in most cases. Most lesions are intradural (10%), although they can also grow extradural (10-15%). The typical symptoms of schwannoma are radicular pain, which is eventually followed by motor deficits.<sup>1,6</sup>

The clinical presentation of spinal cord tumors depends on the level of the tumor. The most common clinical symptom in adults is worsening pain in a semi-recumbent position or at night. In IDEM, neurological symptoms often occur over time due to the compressed spinal cord and depend on the tumor location. IDEM tumors often grow anteriorly or posterolaterally; therefore, Brown-Sequard Syndrome is often experienced by the patients.<sup>1</sup>

Surgical intervention for intradural extramedullary tumors aims to control the origin of the tumor. Direct tumor control after opening the dura can be performed in the posterior or lateral tumor matrix without mobilizing the spinal cord.<sup>1</sup> Surgical intervention can provide significant symptom improvement in patients

with IDEM tumors. The aim of this study is to provide an overview of the clinical improvement of patients with IDEM tumors after undergoing surgical management in Dr. Moewardi General Hospital from 2019-2022.

## 2. Methods

This retrospective descriptive study was conducted in Dr. Moewardi General Hospital. A total of 44 patients who underwent surgery for IDEM spinal cord tumors in 2019-2022 were included by first obtaining ethical approval (274/II/HREC/2022). The studied variables include age, pathological diagnosis, surgical intervention, and postoperative follow-up. Surgical intervention was conducted in accordance with the symptoms, physical examination, and radiological findings in the MRI, as in the examples in Figures 1 and 2.

The inclusion criteria were all patients with IDEM tumors from 2019-2022 who were admitted to Dr. Moewardi General Hospital. Patients who refused surgery, received conservative treatment, and did not return for a follow-up, were excluded from the study. Data analysis was performed descriptively by observing the distribution of the patients. Data were presented in frequency and percentage in tables.

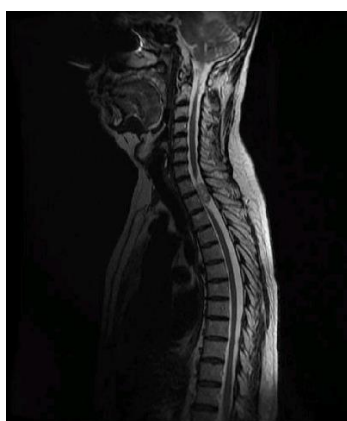


Figure 1. T2 MRI sagittal view showing tumor at Th 1-2 level.



Figure 2. Myelography view showing filling defect at Th 1-2 level.

### 3. Results

A total of 44 subjects were included in the study. The age range was 9-68 years, with a mean age of 45 years. Most subjects were in the age group of 40-60 years (52.3%) and were women (63.6%). Based on the pathology result, the most commonly found in this study was schwannoma (45.5%), followed by meningioma WHO Grade I (25%). The subjects' characteristics are shown in Table 1. The majority of patients who have undergone surgery show improvement in clinical symptoms (86.4%), as summarized in Table 2.

Table 3 shows the distribution based on gender. In this study, 36.4% of the subjects were men, while

63.6% were women. In both men and women, most subjects had schwannoma (56.25% and 39.3%, respectively), followed by meningioma WHO Grade I (25% of each). In this study, we also found one patient with a spinal cord tumor due to metastatic carcinoma suspected to originate from thyroid cancer.

Table 4 shows the distribution based on age group. Most subjects were in the age group of 40-60 years (52.3%). In the age group of <40 years and 40-60 years, most had schwannoma (73.3% and 39.1%, respectively). Meanwhile, in the age group of >60 years, most subjects had meningioma WHO Grade I and psammomatous meningioma WHO Grade I (33.3% of each).

Table 1. Characteristics of the subjects.

Variable	n (%)
<b>Age (years)</b>	
Mean	45
Min-Max	9-68
<40 years	15 (34.1%)
40-60 years	23 (52.3%)
>60 years	6 (13.6%)
<b>Gender</b>	
Men	16 (36.4%)
Women	28 (63.6%)
<b>Pathology</b>	
Schwannoma	20 (45.5%)
Meningioma WHO Grade I	11 (25%)
Psammomatous meningioma, WHO Grade I	6 (13.6%)
Ancient schwannoma	2 (4.5%)
Meningothelial meningioma (WHO Grade I)	1 (2.3%)
Clear cell meningiomas, WHO Grade II	1 (2.3%)
Diffuse B-cell NHL	1 (2.3%)
Transitional Meningioma (WHO Grade I)	1 (2.3%)
Metastasized carcinoma (suspected of thyroid cancer)	1 (2.3%)

Table 2. Distribution based on intervention.

<b>Intervention</b>	<b>n (%)</b>
Laminectomy excision	43 (97.7%)
Laminectomy decompression	1 (2.3%)
<b>Follow-up Result</b>	
No changes	6 (13.6%)
Improved clinical symptoms	38 (86.4%)

Table 3. Distribution based on gender.

<b>Pathology</b>	<b>Men (n=16)</b>	<b>Women (n=28)</b>
Schwannoma	9 (56.25%)	11 (39.3%)
Meningioma WHO Grade I	4 (25%)	7 (25%)
Psammomatous meningioma, WHO Grade I	1 (6.25%)	5 (17.9%)
Ancient schwannoma	1 (6.25%)	1 (3.6%)
Meningothelial meningioma (WHO Grade I)	0	1 (3.6%)
Clear cell meningiomas, WHO Grade II	0	1 (3.6%)
Diffuse B-Cell NHL	0	1 (3.6%)
Transitional meningioma (WHO Grade I)	0	1 (3.6%)
Metastasized carcinoma (suspected of thyroid cancer)	1 (6.25%)	0

Table 4. Distribution based on age group.

<b>Pathology</b>	<b>&lt;40 (n=15)</b>	<b>40-60 (n=23)</b>	<b>&gt;60 (n=6)</b>
Schwannoma	11 (73.3%)	9 (39.1%)	0
Meningioma WHO Grade I	2 (13.3%)	7 (30.4%)	2 (33.3%)
Psammomatous Meningioma, WHO Grade I	1 (6.7%)	3 (13%)	2 (33.3%)
Ancient schwannoma	0	1 (4.3%)	1 (16.7%)
Meningothelial meningioma (WHO Grade I)	0	0	1 (16.7%)
Clear cell meningiomas, WHO Grade II	1 (6.7%)	0	0
Diffuse B-Cell NHL	0	1 (4.3%)	0
Transitional meningioma (WHO Grade I)	0	1 (4.3%)	0
Metastasized carcinoma (suspected of thyroid cancer)	0	1 (4.3%)	0

#### 4. Discussion

Primary spinal cord tumors are ten times less common than intracranial tumors, representing 2-4% of all primary CNS tumors. Intradural extramedullary tumors (IDEM) are generally benign neoplasms that arise in the spinal canal. This tumor accounts for two-thirds of the primary spinal tumor and 15% of tumors affecting the CNS. Of all primary spinal tumors, IDEM accounts for 80% in adults and 65% in pediatric patients.<sup>7,8</sup> Another study in Ukraine reported that IDEM accounts for approximately 60% of all spinal tumors.<sup>9</sup> The most frequently found tumors are schwannoma, meningioma, and neurofibroma.<sup>7,8</sup>

In our study, most subjects were 40-60 years old (52.3%), with a mean age of 45 and a range of 9-68 years. A study by Confano et al. also found a similar result with a mean age of 58 years (range 18-88 years). Fernandes et al. also reported a similar result in which most subjects were in the age group of 40-60 years.

IDEM tumor, especially schwannoma, is known to have a peak incidence between the fourth to sixth decade.<sup>2,7,8,10</sup> On the other hand, meningioma is known to occur more frequently in the fifth and seventh decade of life. The median age at diagnosis is 66 years old. Statistically, the incidence rate in patients aged 40+ years is 18.69/100,000, and in ages 0-19 years, it is 0.16/100,000. Meningiomas of CNS tumors in patients aged 40+ years, age 15-39 years, and age 0-14 years, respectively found in 43.6%, 15.6%, and 1.7% of cases.<sup>11</sup> Our result is in line with the theory, in which most subjects in the age group of >60 years tend to have meningioma instead of schwannoma.<sup>8</sup>

It has been suggested that there is a strong predilection for women, especially in meningioma WHO Grade I. Based on gender, most subjects in our study were women (63.6%). A similar result was reported by Cofano et al., in which a total of 64.7% of

the subjects were women. A study by Narayan et al. also showed women's dominance compared to men's (54.3% vs. 45.75%). This result is in line with the theory of IDEM tumor predilection, in which most patients are women. This is potentially due to the effect of estrogen, although the mechanism is still unclear.<sup>2,5,7,10</sup>

Based on the type, most subjects in this study had schwannoma (45.5%), followed by meningioma (25%). A similar result is also found in a study by Cofano et al., in which most subjects had schwannoma (43.6% and meningioma (37.6%). Another study by Narayan et al. also reported schwannoma as the most frequent finding (34.3%), followed by meningioma (25.7%). Schwannoma is a benign tumor arising from the dorsal sensory roots. This tumor is classified as a nerve sheath tumor (NST) besides neurofibroma and accounts for 23-25% of intradural spinal cord tumors in adults and approximately 14% in pediatric patients.<sup>2,7</sup> The majority of the schwannomas are sporadic. Schwannoma findings in specific syndromes, such as neurofibromatosis type 2, schwannomatosis, and the Carney complex, may have a possible genetic etiology. Genetic studies show that the NF2 gene on chromosome 22 encodes for the merlin protein (schwannomin) and plays an essential role in sporadic and syndromic schwannoma development. Besides, especially in spinal schwannomas, it can have SMARCB1 mutations and inactivation.<sup>12,13</sup> On the other hand, meningioma is a tumor arising from the meningotheial arachnoid cap cells embedded in the dura near the root sleeve. Spinal meningioma accounts for up to 50% of all intradural spinal neoplasms and is known as one of the most frequent spinal tumors in adults. Meningiomas are typically slow-growing tumors. For asymptomatic meningiomas, a linear growth rate of 2-4 mm/year. The loss of chromosome 22q, where the neurofibromatosis type 2 (NF2) gene is located, is also the most common chromosomal abnormality of meningiomas. Aside from the loss of 22q, grade I meningiomas do not display consistent alterations and are typically stable at the cytogenetic level.<sup>14,15</sup>

Moreover, the most common meningioma types are psammomatous, meningothelial, and transitional;<sup>8</sup> in accordance with this statement, our result also showed that 13.6% of our subjects had a psammomatous meningioma.<sup>8</sup>

Currently, there are various surgical approaches for IDEM tumors, including anterior, anterolateral, posterior, and a combination of anterior and posterior approaches. Based on the intervention, all subjects in our study had a laminectomy. A similar result has also been reported by Narayan et al., in which 71.4% of patients underwent laminectomy with a posterior approach, and 94.3% underwent total laminectomy with tumor excision with microsurgery.<sup>2,16</sup> Laminectomy is still regarded as the best treatment for spinal intradural extramedullary tumors, especially schwannomas. Spinal deformity caused by iatrogenic spinal instability due to total laminectomy without internal fixation may damage the posterior column structure, including the bony structure of the posterior ligament complex. Standard surgery usually includes laminectomy for sufficient exposure of the tumor, with facetectomy needed in some cases. Instrumented fixation can be imperative to keep spinal stability and decrease postoperative complications. But the contributing factors related to the need for instrumented fixation during tumor resection remain elusive.<sup>17</sup> Previous studies by Sebai et al., a total of 56 patients, conducted logistic regression analysis and found that total facetectomy and cervicothoracic involvement are strongly associated with the need for instrumented fixation during an intraspinal tumor resection surgery.<sup>18</sup>

In the postoperative follow-up, most subjects in our study reported clinical symptom improvement (86.4%). The improvement in clinical symptoms, mainly due to the compression of the central nervous system by the tumor, has been removed so that nerve function can improve. Complete resection of the IDEM lesion seems desirable to decompress the neural elements and relieve the compressive pathology, but a complete resection may be unnecessary. Patients exhibit some subjective improvement in pain relief and

or motor power post-operatively, even when simple debulking or sub-total resection is performed. In previous literature, tumor resection was reported as en block, whereas others convey adherence of the tumor to neural elements precluding subtotal resection. In a palliation pain control strategy, transection of the affected nerve rootlets if metastatic disease is firmly adhered, precluding complete resection.<sup>19</sup> A study by Narayan et al., who conducted a one-year follow-up, also reported that most subjects (77.14%) reported a good outcome with an improved Frankel score at the end of the follow-up period. Another study by Meng et al. in 2022 also reported a significant difference between the Japanese Orthopaedic Association (JOA) score and Nurick grade in patients before and after laminectomy. Furthermore, another study by Govind et al., who conducted one-month and six-months postoperative follow-ups, also showed that most subjects had an excellent outcome (37.3% and 61.2%, respectively).<sup>2,16,20</sup>

There are several limitations to this study. We did not record data on the patient's initial symptoms; therefore, we could not compare the improvement between the initial presenting and postoperative symptoms. Moreover, we did not use an objective scoring system to evaluate postoperative functional outcomes. Future studies should assess postoperative functional outcomes using an objective and valid scoring system. In addition, more samples are necessary to provide more significant data.

## 5. Conclusion

Intradural extramedullary spinal tumor mostly occurs in women aged 40-60. Most IDEM tumors are schwannoma and meningioma. Surgical intervention of laminectomy can provide good outcomes and improve most patients' clinical symptoms.

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